SAPIENZA
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MEMORY, EMOTION, COGNITION AND WAVES:

neuroscientific navigation through hearing and deafness

Thesis for the Degree of Doctor of Philosophy in
Clinical Experimental Neuroscience and Psychiatry

by
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Cycle XXXV

A chi è capace di ascoltare,
anche senza sentire

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INTRODUCTION

This introduction refers to long and intense years of activity in the field of auditory cognitive neuroscience, years during which there have been many wanderings in the sea of research. Starting from a small experimental question, the *working memory* (WM), I have travelled different routes, some of which have led me to unexpected islands of knowledge, which I would never have thought I would be able to visit in years gone by. Moreover, the research question itself has, so to speak, ancient origins in me. Allow me here, therefore, to reactivate a childhood memory. When I was a student at the conservatory, practising on the flute in Ludwig Van Beethoven's *Serenade*, repeating several passages while trying to play a measure correctly, I wondered if there existed and what was the cerebral mechanism that enabled me, after several repetitions, to acquire the difficult passages in my memory and to be intensely moved when listening both to such melodies played by me as well as to other compositions. Evidently, notes are just black dots on five lines if multisensory memory (sight, hearing, touch for finger articulation) does not get to work, as plainly, emotion does not reside in black dots or individual life events but in the ability of our sensory systems – specifically hearing – to transmit a simple wave oscillating in the air to the stage of our brain.

And it was from this *sound wave* that I started, wondering how its processing could cause substantial differences in such fundamental capacities for healthy development as WM and *emotion processing* in children and adults. Furthermore, such differences are exacerbated in people who, due to sensory hearing deficits, use the *cochlear implant* (CI), a bionic ear resulting from the intense synergy between medicine and research, which, perhaps, Beethoven himself in his deaf years would not have hesitated to use had he had the opportunity.

However, my starting question was what brain mechanisms underlie these abilities: what methodology to study and apply to answer my childish question?

To find an answer, I turned to other *electrical waves* that develop from neurons and can be acquired and analysed through *electroencephalography* (EEG) a technique widely used for clinical diagnostic purposes in medical science. Therefore, considering the objective evidence based on behavioural and psychological data showing how children and adults manifest difficulties in specific cognitive and emotional functions, I went beyond the boundary of this evidence by asking myself about the *brain* source, thus searching for patterns of neurophysiological activations underlying such discrepancies between hearing impaired and normal hearing people.

The main objective of my research work was, therefore, to use the EEG technique to try to identify possible neurophysiological markers, patterns of performance and deficits in cognitive and emotional tasks in different *sensorial modalities* (auditory and visual). Research was carried in groups of children and adults with CIs and to investigate the scientific validity of this neuroimaging technique in such a sophisticated and much interesting clinical population. The possibility of having access, on the one hand, to a unique experimental sample and, on the other hand, to modern electroencephalographic signal acquisition systems, allowed me to elaborate several scientific questions and to design and realise two projects on WM. The first 'Audio-Video EEG correlates of WM in children CI users' focused on auditory and visual verbal working memory, a second 'WHEMORCHING'[1], focused on emotional WM. Of the former, two works published in peer-reviewed journals are reported in this thesis, and one is currently underwriting. Of the second the data acquired are presently under analysis. Participation in the 'Music listening in noise' project on the other hand, allowed me to investigate the neurophysiological correlates of the perception of a stimulus as complex and compelling as *music* and the ability to recognise *emotion* according to the sensorial modality of presentation (visual, auditory) in people with and without CI. A published work is proposed for each of the investigations. Furthermore, again using emotionally charged but negative stimuli, such as those related to substance addiction, I verified the effectiveness of specific *neurophysiological indices*, in one proposed paper. Moreover, through two works focused on *attention*, a cognitive capacity closely correlated with WM, I investigated *psycho-cognitive* patterns in children and elderly CI users.

But, Odysseus teaches us, even when the route seems plotted and the wind seems favourable to smooth sailing towards a safe harbour, Aeolus and Neptune can upset our path, creating terrible and unexpected waves. And the unexpected *wave* in my navigation was that of the Covid 19 dramatic pandemic, which forced the whole world into long quarantine periods, during which it was impossible to carry out contact research as the electroencephalographic setup required.

However, the *pandemic wave*, despite being overwhelming and very complicated to manage while remaining firmly at the helm, offered me the opportunity to cultivate further research questions focused on how normal and hearing-impaired students struggling with *online education*, were experiencing the quarantine isolation period. This question gave rise to two projects, one COCLOVID[2], concerning the school and psychological well-being of hearing-impaired and normal-hearing children and their families. The other, NEURODANTE 2.0, concerning the expressive emotional response to poetry, read and listened, by normal-hearing university students. Moreover, I wrote a chapter in an Italian published book, on the effects of pandemic isolation on children and adolescents.

With the main aim of understanding how far brain waves picked up by EEG can provide answers on cognitive and emotional dysfunctionality in subjects with and without auditory sensory deficits, this thesis is structured in three parts.

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[1] Neurophysiological CHaracteristics of an EMOtional auditory/visual WoRkING memory task in Children with and without Hearing loss

[2] evaLuation of Online Didactic during cOVID pandemiC in Deaf children with or without COCLhear implants and parents

The first part offers a background that provides the anatomical, clinical, and psycho-cognitive context where the scientific questions are entrenched. Therefore, the background will offer firstly a concise overview of auditory stimulus processing, from the uptake performed by the outer ear to processing in the cerebral cortex, with a focus on auditory disorders and the CI. Next, the specific cognitive experimental variables investigated, such as WM, attention and emotions, will be discussed, with an in-depth look at the main behavioural and neurophysiological scientific evidence in people with and without hearing loss.

The second part explains electroencephalography, the main method used to investigate in depth the principal scientific questions, delving into the reasons that led me to use it, its characteristics, and the main stages of its application in experiments.

All the ten scientific questions (from A to L) will be set out in the third part of this thesis, where they will be answered experimentally (ten published works conducted in the laboratory and remotely during pandemic). Schematically, the ten original investigations will cover specifically the following.

The first work will aim to identify a neurophysiological benchmark in the different sensorial modalities in the processing of verbal WM; the second will investigate the differences of *visual* and *auditory* verbal WM between normal hearing and unilaterally cochlear implanted children.

The third work will evaluate the lateralisation of *emotional vocal stimulus* recognition ability in children with unilateral CI. *Music* listened to in presence of different noise background will be the subject of the fourth work, which will assess EEG indices of pleasantness and mental fatigue. The fifth work will evaluate the effectiveness of *neurophysiological indices* analysing emotional responses in another clinical sample. The sixth and the seventh will investigate *attention* in children and adults with cochlear implants using a neuropsychological methodology. Finally, the last three works will investigate the *effects of isolation* during the covid 19 pandemic. In the eighth work the *school well-being* of scholars with and without hearing impairment and their families during online education will be investigated while the ninth will delve theoretically into *online education*, in a chapter of a book written with other clinical colleagues during the pandemic. Finally, the last work will explore the *emotional response to poetry* enjoyed through listening and reading by university students.

INTRODUZIONE

L'introduzione al presente lavoro si basa su lunghi ed intensi anni di attività svolti nell'ambito delle neuroscienze cognitive uditive. Anni durante i quali diverse sono state le peregrinazioni nel mare della ricerca. Partendo da un piccolo quesito sperimentale, la *memoria di lavoro*, ho percorso diverse rotte, alcune delle quali mi hanno condotta in isole di conoscenza inaspettate, che negli anni passati, non avrei mai pensato di riuscire a visitare. Ed è lo stesso quesito di ricerca ad avere in me origini, per così dire, antiche. Mi permetto in questa sede, pertanto, di riattivare un ricordo di infanzia. Quando studentessa di conservatorio, esercitandomi al flauto sulla *Serenade* di Ludwig Van Beethoven, ripetendo diversi passaggi cercando di suonare correttamente una battuta, mi chiedevo se esistesse e quale fosse il meccanismo cerebrale che mi permetteva dopo diverse ripetizioni, di acquisire in memoria i difficoltosi passaggi e di emozionarmi intensamente all'ascolto sia di tali melodie da me suonate, sia durante l'ascolto di altre composizioni. Evidentemente, le note sono solo punti neri su cinque linee se la memoria multisensoriale (vista, udito, tatto per l'articolazione delle dita) non si mette al lavoro, come evidentemente l'emozione non risiede nei punti neri o nei singoli eventi della vita, ma nella capacità che hanno i nostri sistemi sensoriali—nello specifico l'udito— di trasmettere una semplice onda oscillante dall'aria al palcoscenico del nostro cervello.

E proprio da questa *onda sonora* sono partita chiedendomi come la sua elaborazione, potesse essere la causa di differenze sostanziali in capacità fondamentali per uno sviluppo sano quali la memoria di lavoro ed il processamento delle *emozioni* in bambini ed adulti. E tali differenze di fatto si riscontrano acutizzate nelle persone che, a causa di deficit uditivi sensoriali, utilizzano l'*impianto cocleare* (IC), un orecchio bioinico frutto dell'intensa sinergia tra medicina e ricerca, che forse, lo stesso Beethoven nei suoi anni di sordità non avrebbe esitato ad utilizzare se ne avesse avuto l'opportunità.

Tuttavia, la mia domanda di partenza era quali fossero i meccanismi del cervello sottostanti queste abilità: quale metodologia, pertanto, studiare ed applicare per rispondere alla mia infantile domanda?

Per trovare una risposta ad essa, mi sono rivolta ad altre onde, le onde elettriche che sviluppandosi dai neuroni, permettono di essere acquisite ed analizzate attraverso l'*elettroencefalografia* (EEG), tecnica ampiamente utilizzata a fini clinici-diagnostici nella scienza medica.

Pertanto, considerando le obiettive evidenze basate su dati comportamentali e psicologici mostranti come bambini ed adulti manifestino difficoltà in specifiche funzioni cognitive ed emozionali, mi sono spinta oltre il confine di queste evidenze chiedendomene la fonte cerebrale, ricercando dunque *patterns* di attivazioni neurofisiologiche sottostanti a tali discrepanze tra ipoudenti e normoudenti.

L'obiettivo principale del mio lavoro di ricerca è stato dunque, quello di utilizzare l'EEG per provare ad identificare eventuali *pattern* neurofisiologici, *marker* dei deficit delle *performance* a compiti cognitivi ed emozionali in diverse *modalità sensoriali* (uditiva, visiva), in gruppi clinici di bambini ed adulti portatori di IC e valutare la solidità scientifica di tale tecnica di neuroimmagine in tanto sofisticata ed affascinante popolazione clinica. La possibilità di aver accesso da un lato ad un campione sperimentale unico e dall'altro a moderni sistemi di acquisizione del segnale elettroencefalografico, mi ha permesso di elaborare diverse domande di ricerca e di ideare e realizzare due progetti sulla memoria di lavoro, il primo '*Audio-Video EEG correlates of WM in children CI users*' focalizzato sulla memoria di lavoro verbale uditiva e visiva, un secondo WHEMORCHING[3], focalizzato sulla memoria di lavoro emozionale. Del primo nel presente lavoro sono riportati due studi pubblicati su riviste scientifiche internazionali ed uno è in fase di scrittura. Del secondo, si è in fase di analisi dei dati acquisiti. La partecipazione al progetto '*Music listening in noise*' mi ha permesso invece di approfondire i correlati neurofisiologici della percezione di uno stimolo tanto complesso quanto efficace come quello *musicale* nonché la capacità di riconoscimento dell'emozione in base alla *modalità sensoriale* di presentazione (visiva, uditiva) in persone portatrici e non di IC. Inoltre, sempre nell'attivazione per stimoli emozionalmente carichi, ma negativamente quali quelli connessi alla dipendenza da sostanza, in uno studio ho verificato l'efficacia di determinati *indici neurofisiologici*. Attraverso due lavori focalizzati sull'*attenzione*, capacità cognitiva strettamente correlata con la WM, ho avuto modo di approfondire i *pattern* psico-cognitivi in bambini ed anziani portatori di impianto cocleare.

Ma, Odisseo ce lo insegna, anche quando la rotta sembra tracciata ed il vento pare favorevole ad una regolare navigazione verso un porto sicuro, Eolo e Nettuno possono scompaginare il nostro cammino, creando terribili ed inaspettate onde. E l'*onda* inaspettata nella mia navigazione è stata quella della drammatica Pandemia da Covid 19, che ha costretto il mondo intero a lunghi periodi di quarantena, durante i quali, è stato chiaramente impossibile effettuare ricerca a contatto con le persone come richiede il set-up elettroencefalografico.

Tuttavia, l'*onda pandemica*, malgrado sia stata travolgente ed a tratti molto complicata da gestire, rimanendo ben salda al timone, mi ha offerto l'opportunità di coltivare ulteriori domande di ricerca focalizzate su come stessero vivendo il periodo di isolamento gli studenti normo ed ipoudenti alle prese con la *didattica online*. E da qui sono nati due studi, uno COCLOVID[4] inerente al *benessere scolastico* e psicologico dei bambini normo ed ipoudenti e le loro famiglie, l'altro, NEURODANTE 2.0, riguardante la *risposta emozionale* espressiva alla poesia letta ed ascoltata in studenti universitari normoudenti, oltre ad un capitolo in un libro pubblicato in Italia, sugli effetti dell'isolamento da pandemia in bambini ed adolescenti.

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[3] Neurophysiological CHaracteristics of an EMOtional auditory/visual WoRkING memory task in Children with and without Hearing loss

[4] Valutazione della Didattica Online nel corso della quarantena per COVID in bambini portatori di Impianti COCLeari e genitori

Quindi, con l’obiettivo principale di comprendere quanto le onde cerebrali captate dall’ EEG possano fornire risposte significative quantitativamente caratterizzate sulle disfunzionalità cognitive ed emozionali in soggetti con e senza deficit sensoriali uditivi, la presente tesi si struttura in tre parti.

La prima parte presenta il *background* che fornisce il contesto anatomico, clinico e psicocognitivo in cui si collocano le domande scientifiche affrontate in questa tesi. Pertanto, il *background* offrirà una sintetica panoramica sul processamento dello stimolo uditivo, dalla captazione effettuata dall’orecchio esterno sino al processamento in corteccia cerebrale con un focus sui disturbi uditivi e l’impianto cocleare. Successivamente verranno esposte le variabili sperimentali specifiche oggetto degli studi, quali la memoria di lavoro, l’attenzione e le emozioni, approfondendo le principali evidenze scientifiche comportamentali e neurofisiologiche in persone con e senza ipoacusia. La seconda parte illustrerà l'elettroencefalografia, il metodo utilizzato per indagare le principali questioni scientifiche, approfondendo le ragioni che mi hanno spinto a utilizzarla, le sue caratteristiche e le principali fasi della sua applicazione in ambito sperimentale.

Tutte le dieci domande scientifiche (dalla A alla L) saranno esposte nella terza parte della tesi, dove troveranno risposta nei dieci lavori pubblicati. Schematicamente, le originali indagini scientifiche riguarderanno nello specifico quanto segue.

Il primo lavoro mirerà a identificare un *benchmark* neurofisiologico nelle diverse modalità sensoriali di elaborazione della memoria di lavoro verbale in bambini normoudenti mentre il secondo indagherà le differenze nella memoria di lavoro verbale visiva e uditiva tra bambini normoudenti e bambini con IC unilaterale.

Il terzo lavoro valuterà la lateralizzazione della capacità di riconoscimento degli *stimoli emotivi vocali* nei bambini con IC unilaterale. La *musica* ascoltata in diverse condizioni di rumore sarà oggetto del quarto lavoro, che valuterà gli indici EEG di piacevolezza e fatica mentale in adulti con IC unilaterale. Il quinto studio valuterà l'efficacia degli *indici neurofisiologici* che analizzano le risposte emotive in un diverso campione clinico. Il sesto ed il settimo studio analizzeranno l'*attenzione* in bambini e adulti con impianto cocleare utilizzando metodologie neuropsicologiche. Infine, gli ultimi tre lavori indagheranno gli *effetti dell'isolamento* durante la pandemia della Covid 19. Nell’ottavo, si indagherà il *benessere scolastico* di bambini con e senza deficit uditivi e delle loro famiglie durante la didattica online mentre il nono approfondirà dal punto di vista teorico la *didattica a distanza* in un capitolo di un libro scritto coralmente con altri colleghi clinici durante la pandemia. Infine, l’ultimo lavoro esplorerà la *risposta emozionale alla poesia* fruita attraverso l'ascolto e la lettura da parte di studenti universitari.

I. BACKGROUND ON AUDITORY, COGNITIVE AND EMOTIONAL *WAVES*

a. AUDITORY WAVES

I. 1. Hearing

Hearing perception in humans is one of the most essential sensory modalities because of its role in understanding and producing speech. The ability to distinguish between a wide range of lighter, louder, simple and complex sounds, allows for the complete enrichment of our existences that begins in the womb, continues in school, evolves into the adult world and deeps into silent elderly solitude. Hearing sounds allow us to experience and thus construct ourselves, from the first notes of life to the complex symphony of our personalities.

Sound is a mechanical disturbance from a state of equilibrium, consisting of compressions and rarefactions that propagate through the air at a speed of about 340 m/s (figure 1). Observing a soprano at the Opera as she tries her hand at the highest notes or when we try to shout to produce the pressure changes in the air necessary for the genesis of sounds, it is necessary to perform work on the air through our vocal apparatus or by another source of sound natural or man-made (e.g., wind, flowing streams, avalanches, volcanoes, musical instrument, trains, fans etc.). The sound thus generated transmits acoustic energy through the air. In order to hear, each of the two ears must concentrate this mechanical energy and transmit it to the receptive organ of the ear, which converts it into an electrical signal that the nervous system can analyse. These tasks correspond to the respective functions of the outer ear, middle ear, and inner ear. The details of this incredible, almost instantaneous, energy transformation will follow.

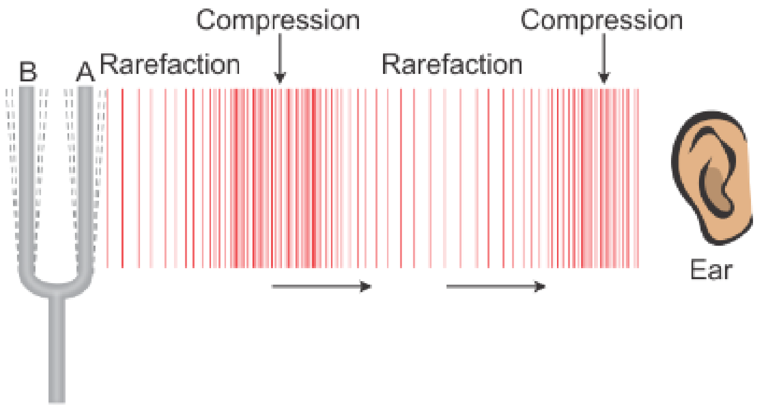

Figure 1: A vibrating tuning fork disturbs the air, producing alternating high-pressure regions (compressions) and low-pressure regions (rarefactions), which form sound waves. (Source: toppr.com, 2023).

Like a parabolic antenna collects electromagnetic radiation, the external part of the ear, or pinna, collects the air pressure variations that carry sound by channelling them along the external auditory meatus or ear canal. The external meatus ends at the tympanic membrane, a thin diaphragm approximately 9 mm in diameter. The mechanical energy from the sounds in the air passes through the middle ear via the coordinated movement of the ossicular chain: malleus, incus, and stapes (figure 2). The base of the malleus is attached to the tympanic membrane, and a ligament connects the other end to the incus, which in turn is connected to the stapes. The end of the stapes, known as its footplate, presses onto the oval window, dug into the bony surface of the snail-shaped, fluid-filled cochlea, the inner ear (Hudspeth, 2019) (figure 2).

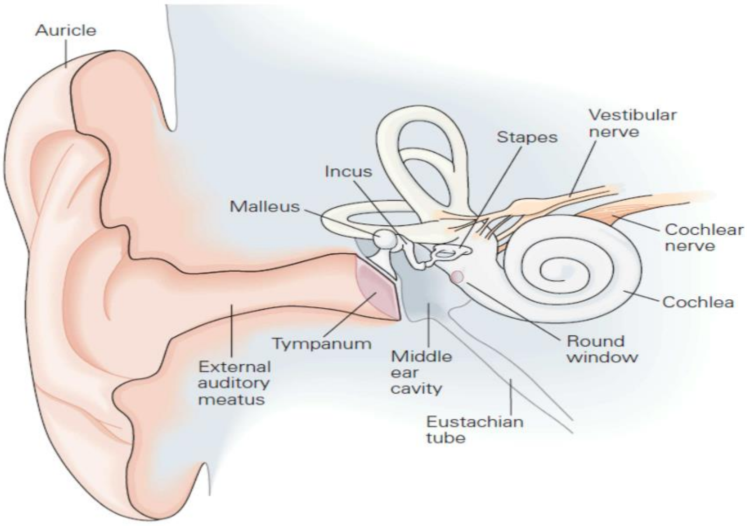

Figure 2. The structure of the human ear. The external ear, especially the prominent auricle, focuses sound into the external auditory meatus. Alternating increases and decreases in air pressure vibrate the tympanum. Three tiny, linked bones convey these vibrations across the air-filled middle ear: the malleus, the incus, and the stapes. The vibration of the stapes stimulates the cochlea, the hearing organ of the inner ear. (Source: Kandel et al., 2013).

The inner ear, or cochlea (from the Greek word κοχλίας= kokhlías, "spiral, shell or snail)[5], is the size of a pea with a transverse diameter of about 9 mm, is covered by a thin bony lamina and is hidden deep within the dense structure of the temporal bone. The interior of the cochlea, when viewed in a transverse section (figure 3), consists of three fluid-filled compartments called scales: the *tympanic* scale, the *vestibular* scale, and the *media* scale (cochlear canal) (Reichenbach and Hudspeth, 2014).

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[5] Collins Greek-English dictionary: (Collins, 2023).

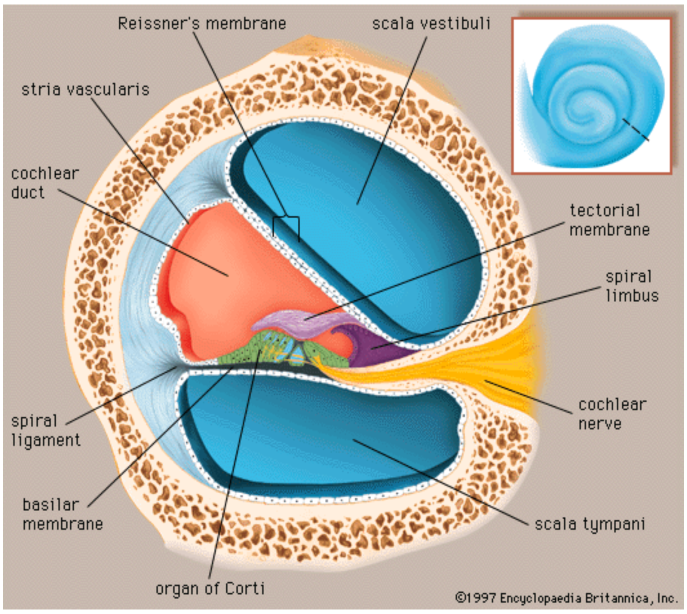


Figure 3. Structure of the cochlea, inner ear. A cross-section through one of the turns of the cochlea (inset) showing the scala tympani and scala vestibuli, which contain perilymph, and the cochlear duct, which is filled with endolymph. (Source: Encyclopædia Britannica, Inc, s.d.).

The fluid composition in the vestibular and tympanic scales is the same, sodium-rich, extracellular fluid. The fluid in the media scale has a different ionic composition, rich in potassium, similar to the composition of the intracellular fluid. The basilar and Reissner's membranes separate the fluid with a high potassium concentration (endolymph) from the fluid with a high sodium concentration (perilymph) (Møller, 2018a).

The mechanical properties of the basilar membrane constitute the key element of cochlear function (Ruggiero, 1992). When the stapes moves due to a sound reaching the tympanic membrane, it sets the fluid in the cochlea in motion, resulting in a specific movement of the basilar membrane known as a 'travelling wave'. Since mechanical stimulation is applied to the base of the cochlea as a pressure difference between the vestibular and tympanic scales, the travelling wave propagates from the base of the cochlea towards the apex. Each wave reaches its maximum amplitude at a particular location that depends on the frequency of the stimulus and then rapidly decreases as it travels towards the apex of the cochlea. This reminds of the marine waves whose crest reaches a maximum height as it approaches the beach and then it quickly breaks and vanishes. Although the metaphor is certainly suggestive, it should be emphasised that the physical basis of the two phenomena is entirely different. The energy of the sea wave depends on the momentum of a mass of water moved by the wind (Veerabhadrappa et al., 2022). On the other hand, the movement of the basilar membrane comes from the movement of the fluid masses above and below the membrane. These fluid masses, in turn, are pushed up and down by the energy produced by the piston-like movement of the stapes at the oval window. Variations in the mechanical properties of the basilar membrane explain why the membrane is tuned to different frequencies in hertz (Hz) at each point along its course. The result is that the basilar membrane's elasticity decreases with distance from the base. A specific frequency of vibration thus preferentially resonates at a specific distance from the base, as elucidated in the 1940s by Georg von Békésy (1960). For this work, he received the Nobel Prize in physiology or medicine in 1961. To compare it to a musical instrument, the basilar membrane is played by acoustic energy, much like a xylophone, as shown in figure 4.

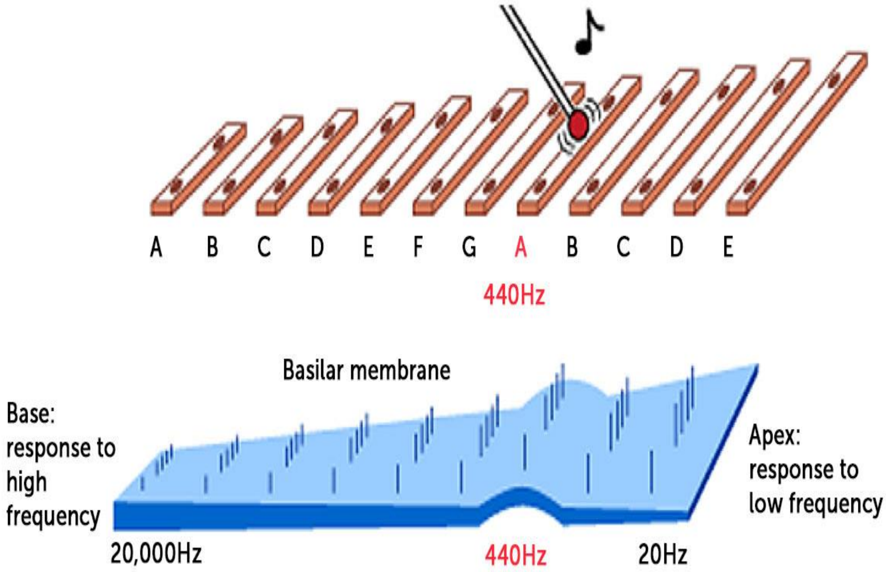


Figure 4. Representation of the similarity of frequency distribution between cochlea and xylophone. (Source: Brain Hq, s.d.).

It was the German physiologist Hermann Helmholtz (1821-1894) who understood the way the basilar membrane operates as a frequency analyser. Each point on the basilar membrane is tuned to a specific frequency. In humans, at the apex of the cochlea, the basilar membrane responds to the lowest auditory frequencies (20 Hz) and at its base to the highest frequencies (20kHz). This fixed array of frequency sensitivity down the length of the cochlea is known as *tonotopy* or frequency-to-place mapping. It is essential and fascinating to remember how the same tonotopy present in the cochlea is mirrored centrally in the primary auditory cortex (Romani et al., 1982) (figure 5), leading to the reflection of how human listening is a function as complex as it is regular and made up of elements that refrain. Yet, as in the most complex compositions, the basic chords return with regularity, to harmonise the apparent chaos.

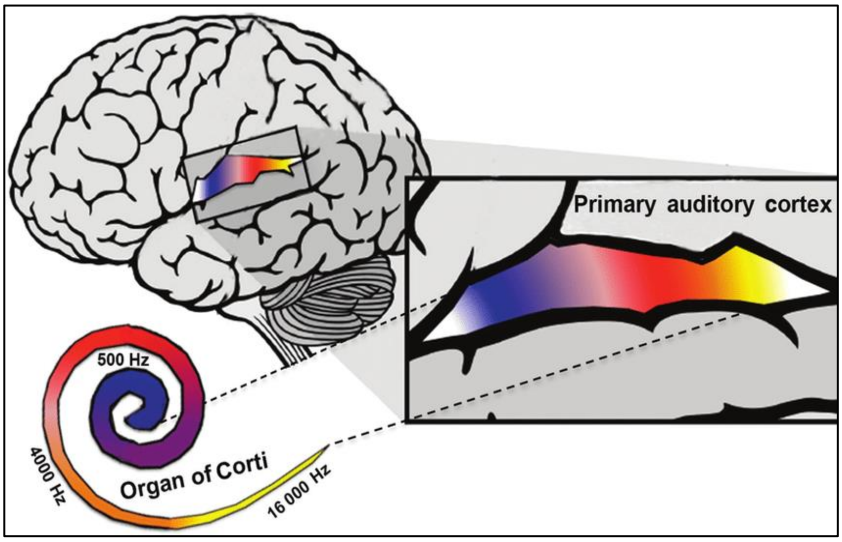


Figure 5. Tonotopy in the auditory system. Different regions of the basilar membrane in the organ of Corti, or spiral organ, located in the cochlea, vibrate at different frequencies (500–16 000 Hz). This topographical mapping, or tonotopy, is maintained along the stations of the auditory pathway all the way to the primary auditory cortex in the temporal lobe. Therefore, as in the organ of Corti, in the auditory cortex, neighbouring neurons respond to nearby frequencies, shown as different colours. (Source: Perrone-Capano et al., 2017).

Corti's organ (figure 6) is the cochlea site, where mechanoelectrical transduction takes place, and is the most mechanically complex structure in the human body (Reichenbach and Hudspeth, 2014). It consists of a strip of epithelial tissue covering the basilar membrane's elastic structure. It contains approximately 16 000 hair cells (the auditory sensory cells, a kind of mechanoreceptor and activator) innervated by 30 000 afferent nerve fibres that convey the information to the central nervous system by joining the fibres of the eighth pair of cranial nerves. The organ of the ear comprises a wide variety of cell types, of many of them we do not know the function of (scientific research still has much to explore inside the ear!). The hair cells, immersed in the fluid of the media scale, are divided into *internal* and *external*. The former perceive the vibration of the basilar membrane and transfer the electrical stimulation (action potential) to the auditory nerve fibres that terminate on them (Møller, 2018a; Plomp and Smoorenburg, 1970). The outer hair cells react to the deflection of the cilia (stereocilia) by lengthening and shortening in response to the deflection of their stereocilia. In practice, they help the basilar membrane to vibrate by amplifying it, improving the hearing threshold by about 50 deciBel (dB). They are the most vulnerable compared to the inner cycloid cells. The hair cells, stimulated by the wave, transmit the fruits of their speedy work to the central nervous system via the auditory nerve, with which they have synaptic-like connections.

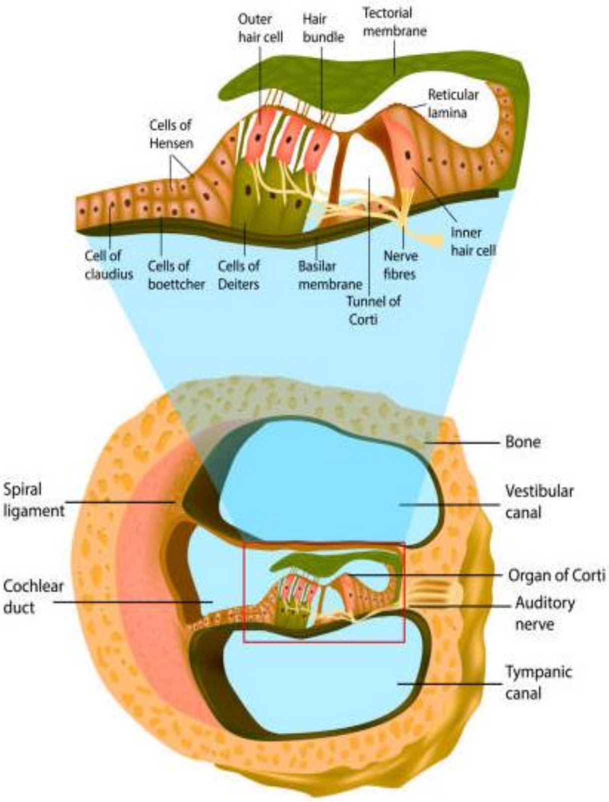


Figure 6. Anatomy of the inner ear. Cross section of a cochlea spiral. Structure of the organ of Corti. (Source: istockphoto.com, 2023).

The central auditory system, which extends from the ear to the brainstem and reaches the cerebral cortex through the midbrain and thalamus, can be compared to a powerful, speedy signal processor, which processes rapidly sound's information from the cochlea, separating different types of sounds. In fact, the auditory system is built for speed. Hair cells transduce stimuli in microseconds, a striking contrast to the tens to hundreds of milliseconds required by photoreceptors and olfactory neurons. In keeping with this intense signalling activity, histochemical staining reveals that the auditory system has the highest metabolic rate in the brain (Hudspeth and Konishi, 2000). The organisation is characterised by two parallel pathways: ascending and descending. Among the *ascending* are the classical pathway (lemniscal or specific) and the non-classical (extra-lemniscal or non-specific) ascending pathway (Moeller and Rollins, 2002) (figure 7). The sensory pathway of audition ascends through three brainstem nuclei; both pathways share the first tract of the ascending auditory system. Since about half of the fibres of the auditory pathways cross the midline while others ascend on the same side of the brain, each ear is represented in both the right and left cortex. For this reason, even when the auditory cortical area of one side is injured by trauma or stroke, the binaural hearing may be little affected. Impaired hearing due to bilateral cortical injury involving both auditory areas has been reported, but it is extremely rare; for a case study see (Narayanan et al., 2017).

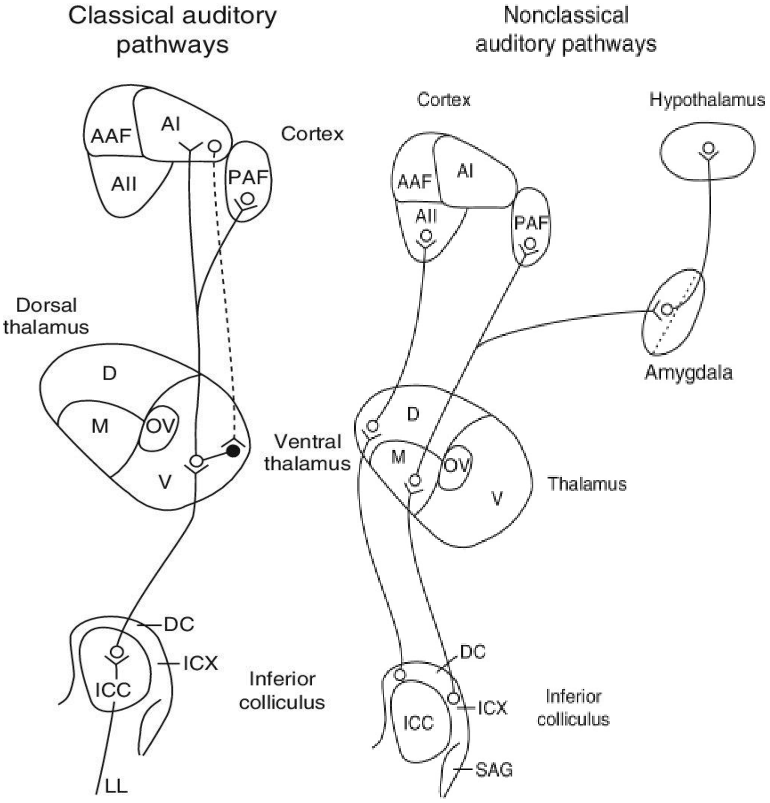


Figure 7. Ascending auditory pathways.
(Left): Schematic drawing of the ascending connections from the external inferior colliculus (ICX) and dorsal cortex (DC) to the thalamic nuclei (MGB) and some of their cortical radiations. M medial (or magnocellular) division of MGB, D dorsal division, V ventral division, OV ovoid part of the MGB. Connections from the MGB to auditory cortical areas and the basolateral nuclei of the amygdala are also shown.
(Right): Schematic drawing of the ascending pathways from the central nucleus of the inferior colliculus (ICC) to the ventral portion of the thalamic nucleus, the medial geniculate body (MGB), and their connections to auditory cortical radiations. Most of the connections have reciprocal descending connections, only one of which is shown in this graph (between AI and the MGB). M medial (or magnocellular) division of MGB, D dorsal division, V ventral division, OV ovoid part of the MGB. (Source: Møller, 2011).

Along the ‘classical’ ascending pathway (figure 7 left), the fibres of the cochlear nerve (eighth pair of cranial nerves) terminate in the cochlear nuclei of the brainstem, which are divided into three parts: ventral anterior and posterior, and dorsal. Each axon of the auditory nerve connects to each part (a basic feature of the parallel processing principle of the auditory nervous system). These nuclei neurons project to the inferior colliculus located in the midbrain. Some cells terminate directly in the inferior colliculus; others first make contact with the superior olivary nucleus and the nuclei of the lateral lemniscus. The superior olivary nucleus begins the process of sound localisation by estimating the time difference and intensity between each ear (Christov et al., 2018). The inferior colliculus (IC) is a midbrain structure that integrates the vast majority of ascending auditory information and projects via the thalamus's medial geniculate nucleus to the cerebral cortex's temporal lobes in the primary auditory cortex located in Heschel’s gyrus. The IC is also a point of convergence for corticofugal (information from the cerebral cortex) input and input originating outside the auditory pathway. Pathophysiological changes in the IC can alter all aspects of auditory perception (for example tinnitus which will be discussed later) and aid in the localization of sound (Berger and Coober, 2015).

The ‘non-classical’ ascending hearing pathway (figure 7 right) is less known than the classical and more complex and mysterious (Moeller and Rollins, 2002). Its nuclei receive input from all sensory systems in addition to the auditory system. The pathway originates from the midbrain in the external nucleus and the dorsal cortex of the inferior colliculus. The non-classical pathway utilises the dorsal and medial parts of the medial geniculate body that project to the secondary and associative cortex, bypassing the primary auditory cortex. In addition, some cells connect to other brain structures, such as the amygdala and hippocampus. These connections already show, at an anatomical-functional level, the close link between hearing and the interaction between memory, and emotions (Phelps, 2004).

It is worth noting here that while all cells in the nuclei of the classical pathway afferent to the primary cortex respond only to the auditory sensory modality, some cells in the nuclei of the non-classical pathway respond to more than one sensory modality, anticipating in the pathway of sound transmission the multimodality and cross-sensory modality that are among the primary goals of the last section of the present work. Moreover, the existence of the non-classical pathway, which transfers auditory information from the dorsal medial component of the thalamus directly to the subcortical areas of the lateral nucleus of the amygdala (Le Doux, 1992), provides less elaborated information than the classical pathway, but more immediate and irrational.

Concerning the descending auditory pathway, this system of circuits, within the auditory system helps to modulate auditory processing at every level (Peterson et al., 2022). The auditory cortex has direct bilateral projections back to the inferior colliculus, the superior olivary complex, and the cochlear nucleus (Schofield and Coomes, 2005; Coomes, Peterson and Schofield, 2007). These circuits contact neurons in these nuclei that project to every level of the central auditory system and to the cochlea (to modulate outer hair cells) within the peripheral auditory system. These descending circuits help to modulate auditory attention based on an individual's relevance, attention, learned behaviours, and emotional state. Such higher-order functions originate from many regions of the brain (e.g., prefrontal cortex, hippocampus, nucleus basalis of Meynert, and limbic circuits) that have either direct and indirect connections with each other and auditory cortex (Forbes and Grafman, 2010; Weinberger, 2007).

As anticipated, the target of ascending auditory information is the auditory cortex.Although the auditory cortex has several subdivisions, a broad distinction can be made between a primary area and secondary, or belt, areas (Purves et al., 2001) (figure 8).

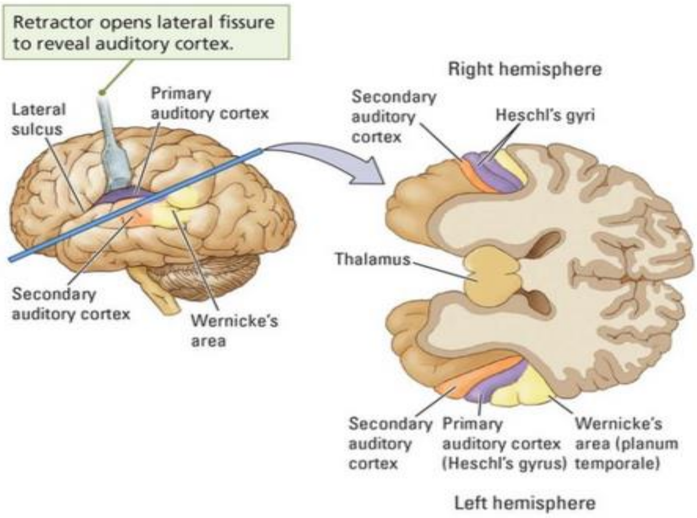


Figure 8. Auditory cortex. The primary auditory cortex, called A1 or PAC, lies within Heschl's gyrus and is surrounded by secondary cortical areas, called A2. Note that Wernicke's area, a region important in comprehending speech, is just posterior to the PAC. (Source: Kolb et al., 2016).

Primary auditory cortex (PAC, A1), located in the depth of the Sylvian fissure (lateral sulcus) where occupies most of the Heschl's gyrus, is cytoarchitecturally defined in the area 41 of Brodmann (Morosan et al., 2011) and secondary (A2) in the adjacent temporal and parietal lobe areas. As anticipated, A1 contains a tonotopic map of the cochlea (figure 9), just as the primary visual cortex and primary somatosensory cortex have topographic maps of their respective sensory epithelium. The preservation of tonotopic (or cochleotopic) (Brewer and Barton, 2016) organisation, one dimension of acoustic feature space, from the basilar membrane to the auditory cortex, allows for a common reference frame in this hierarchically organised sensory system (Kaas, 1997; Wessinger et al., 2001).

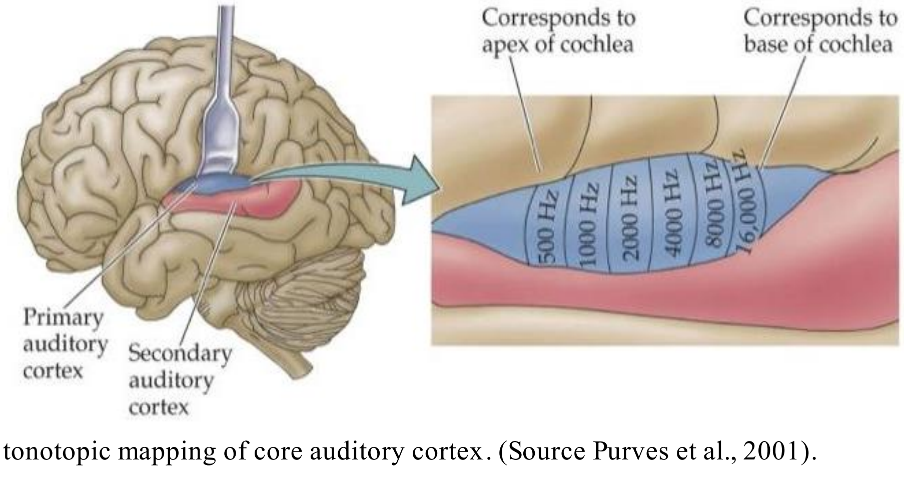


Figure 9. The tonotopic mapping of core auditory cortex. (Source Purves et al., 2001).

The belt areas of the auditory cortex indeed receive more diffuse input from the belt areas of the medial geniculate complex and therefore are less precise in their tonotopic organisation. So, while PAC is essential for fundamental auditory functions such as the recognition of different frequencies and the localisation of sounds as well as playing an essential role in intraspecific communication; the areas surrounding the PAC present a less defined tonotopic organisation and process sounds as complex as language (Purves et al., 2001). It seems likely, therefore, that specific regions of the human auditory cortex are specialised for processing elementary speech sounds as well as other temporally complex acoustical signals, such as music. Indeed, Wernicke's area, which is critical to human language comprehension, lies within the secondary auditory area (Purves et al., 2001).

Hence, the inextricable brain connection between the auditory cortex and language areas emerges. An inextricable connection that, as it will be deepened, is of fundamental importance in hearing-impaired populations. As it will soon be seen, the auditory system is the site of numerous critical periods, including language learning and can turn into plasticity in adults (Oertel and Doupe, 2019).

I. 2. Hearing Loss

Hearing loss, the most common form of human sensory deficit worldwide, (Sheffield and Smith, 2019), consists of the partial or total inability to hear sounds in one or both ears (Bahmad, 2015). Hearing deficit negatively affects well-being and is a major contributor to years lived with disability (James et al., 2018). Over 5% of the world's population requires rehabilitation to address hearing loss (432 million adults and 34 million children). World Health Organization (2023) estimated that by 2050, over 700 million people —or 1 in every 10 people — will have disabling hearing loss.

Impairments anywhere along the auditory pathway, from the external auditory canal to the central nervous system, may result in hearing loss (Nadol, 1993). Congenital hearing loss — hearing loss present at birth — occurs when the ability of the ear to convert the vibratory mechanical energy of sound into the electrical energy of nerve impulses is impaired and is categorised according to the site of the lesion (Korver et al., 2017) (figure 10).

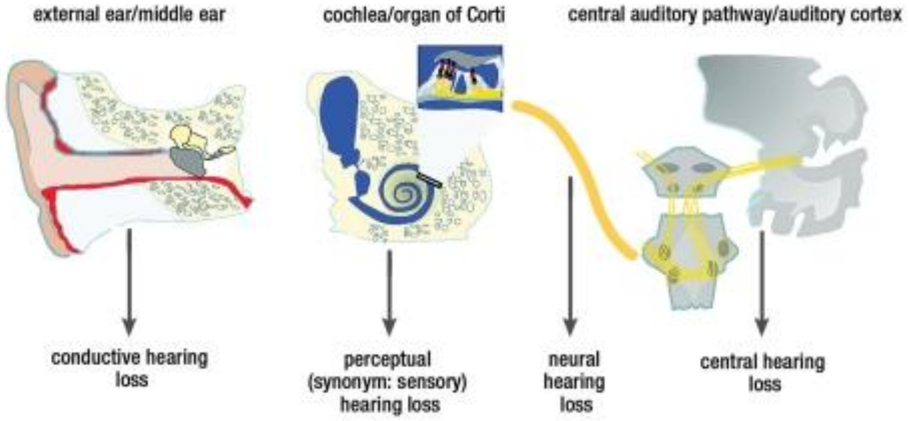


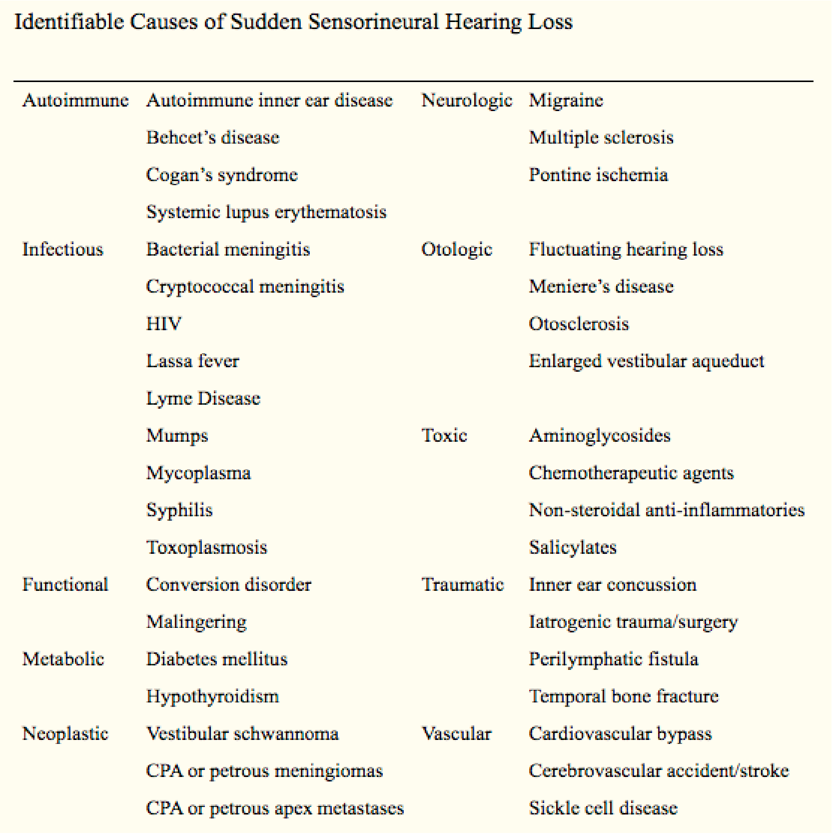
Figure 10. Topographic-functional classification of hearing impairment according to the level of the lesion in the organs of hearing. (Source: Zahnert, 2011).

It is, therefore, possible to distinguish:

i. Transmission (or conduction) hearing loss, linked to malformities, traumatic and often inflammatory alterations of the structures responsible for the mechanical transmission of sound (outer and middle ear) (see Nguyen et al., 2019 for a pictorial review). Congenital causes of conductive hearing loss may be readily apparent on medical, physical examination, such as microtia and aural atresia (Luquetti et al., 2012). More subtle congenital causes of conductive hearing loss, such as ossicular malformations, sclerosis or fixation, can coexist with canal atresia or occur independently. Conductive hearing loss related to eustachian tube dysfunction is commonly encountered in children with Down syndrome, Pierre Robin Sequence (with or without corresponding syndrome), Treacher Collins, FGFR mutations (Apert, Crouzon, and Pfeiffer), Branchio-Oto-Renal, Goldenhar, and Turner syndromes (Bluestone et al., 2014).

ii. Sensorineural hearing loss (SSNHL), where the alteration is localised cranially with respect to the sound conduction apparatus, possibly affecting the cochlea, the auditory nerve (neural hearing loss) or the central auditory pathways (central hearing loss). Sensorineural hearing loss can be further subdivided into sensory hearing loss (when the hair cells are affected), central hearing loss when the cause is located along the central auditory pathway or Auditory Neuropathy Spectrum Disorder (Rapin and Gravel, 2006).

The aetiology is heterogeneous: diagnosis for SSNHL includes, in fact, more than a hundred potential aetiologies (Chau et al., 2010; Mattox and Simmons, 1977), see (Young, 2020) for a recent review. Although, cases with a potentially discoverable aetiology fall into one of several broad categories, including infectious, autoimmune, traumatic, vascular, neoplastic, metabolic, and neurologic (figure 11) it is believed that about 50% of cases of prelingual onset sensorineural hearing loss are due to genetic causes and up to 30% environmental causes.



Identifiable Causes of Sudden Sensorineural Hearing Loss

Autoimmune	Autoimmune inner ear disease	Neurologic	Migraine
	Behcet's disease		Multiple sclerosis
	Cogan's syndrome		Pontine ischemia
	Systemic lupus erythematosis		
Infectious	Bacterial meningitis	Otologic	Fluctuating hearing loss
	Cryptococcal meningitis		Meniere's disease
	HIV		Otosclerosis
	Lassa fever		Enlarged vestibular aqueduct
	Lyme Disease		
	Mumps	Toxic	Aminoglycosides
	Mycoplasma		Chemotherapeutic agents
	Syphilis		Non-steroidal anti-inflammatories
	Toxoplasmosis		Salicylates
Functional	Conversion disorder	Traumatic	Inner ear concussion
	Malingering		Iatrogenic trauma/surgery
Metabolic	Diabetes mellitus		Perilymphatic fistula
	Hypothyroidism		Temporal bone fracture
Neoplastic	Vestibular schwannoma	Vascular	Cardiovascular bypass
	CPA or petrous meningiomas		Cerebrovascular accident/stroke
	CPA or petrous apex metastases		Sickle cell disease

Figure 11. Identifiable Causes of Sudden Sensorineural Hearing Loss. (Source: Kuhn et al., 2011).

The aetiological causes of deafness are heterogeneous, and their early identification is crucial for clinical intervention, especially in children, given its significant, violent impact on linguistic, social, and academic skills (Saraiya and Geller, 2023). Depending on the cause of the deafness, hearing impairments are classified into:

- *Genetic forms*, of which 70% are non-syndromic, i.e., do not associate deafness with other organ or system pathologies, while more than 400 genetic syndromes that include hearing loss have been described (Toriello and Smith, 2004). In fact, about 30% of hearing loss cases in children are classified as syndromic (Angeli et al., 2012), the most frequent are those with autosomal-recessive transmission (85%), including Usher, Pendred and Goldenhar syndromes. On the other hand, autosomal-dominant forms account for only 10%, including Waardenburg, Treacher-Collins-Franceschetti, Brato-Oto-Renale and CHARGE syndromes. The remaining 5% are linked to the X chromosome and mitochondrial DNA (Alport, Norrie, Kearns-Sayre syndromes) (Gorlin et al., 1995).

More than 110 different mutations have been identified. Since the genetics of deafness was not one of the objectives of my doctoral research, I will limit myself to a superficial description of the most widespread mutations, referring to other works for further investigation. However, I think it is important to mention them in a broad perspective, considering importance of the genetic and epigenetic aspects in development and throughout the human life span. To keep in mind that, what observed in my experimental studies, emerges from a complex combination of infinite factors that cannot all be controlled.

So, the most frequent mutation in the majority of Caucasian populations is 35del G mutation and may account for 70% of all GJB2 mutations (Snoeckx et al., 2005). The GJB2 gene encodes connexin 26, a gap junction protein that allows the passage of potassium ions in the inner ear (Angeli et al., 2012). Mutations in the GJB2 gene produce considerable phenotypic variation, and the degree of deafness can vary from mild to profound. Depending on the transmission mechanism/inheritance of the transformation, different forms are recognised: autosomal-dominant; autosomal-recessive; X-liked; mitochondrial. Mutations in SLC26A4 are the second most frequent cause of autosomal recessive no syndromic HL (ARNSHL). The resulting phenotypes include Pendred Syndrome, an autosomal recessive disorder characterised by sensorineural deafness and goitre (Everett et al., 1997).The deafness is congenital and associated with temporal bone abnormalities. Other genes involved in the genesis of deafness are MYO15A, OTOF, ODH23, TMC1, WFS1, MY07A, and COCH.

-*Acquired forms*: they can occur during pregnancy (prenatal), at birth (perinatal) or in the first months/years of life (postnatal). Many deafnesses acquired prenatally, are caused by a group of infectious agents known today as the TORCH complex (Toxoplasmosis, Others, Rubella, Cytomegalovirus, Herpes virus) (Jaan and Rajnik, 2022). In general, the severity of the consequences of the infection depends on the gestational period in which the virus is contracted (Korver et al., 2017). Birth hypoxia, asphyxia and ischemia are often considered major causes of early hearing loss or deafness (perinatal) (Borg, 1997). In fact, the presence of hypoxic-ischaemic encephalopathy has been identified as presenting a significant risk factor for hearing impairment (Anand et al., 1991). Babies of low gestation and birthweight have been reported to be more prone to develop sensorineural hearing loss than term neonates; the prevalence of sensorineural hearing impairment in this population varies from 0 to 4% in reported studies (Marlow et al., 2000). Overall, a prolonged stay in a neonatal intensive care unit (NICU) (especially for incubator noise (Douek et al., 1976) and the duration of artificial ventilation were found to predict best the presence of hearing impairment of *perinatal* origin (Newton, 2001). Hyperbilirubinemia is also found a factor associated with sensorineural hearing loss in NICU graduates and is also associated with auditory neuropathy (Roizen, 2003).

Among *postnatal* hearing loss, the most frequent forms are attributable to infectious, traumatic and toxic causes. In children, hearing loss from ototoxic medications, such as gentamycin, and toxic substances, such as lead, is an infrequent and preventable cause of sensorineural hearing loss and meningitis infections (Stevens et al., 2003). Finally, there is a significant incidence of both sensorineural and conductive hearing loss who suffered head injuries. (Zimmerman et al., 1993).

- *Idiopathic* forms include all cases in which the disorder's aetiology cannot be concretely identified.

- *Presbycusis*: Presbycusis, literally “old hearing” or “elder hearing” (Miller et al., 2005), is the general term applied to age-related hearing loss, includes all conditions that lead to hearing loss in elderly people (i.e. aged 60 years or older, (United Nations, 2019). The disorder is characterised by reduced hearing sensitivity and speech understanding in noisy environments, slowed central processing of acoustic information, and impaired localisation of sound sources. As a result, people with presbycusis have difficulty, proportional to the degree of hearing impairment, in conversation, music appreciation, orientation to alarms, and participation in social activities (Gates and Mills, 2005). It is important to note that 80% of hearing loss cases occur in elderly people (Davis, 1991), and approximately 30% of men and 20% of women in Europe have a hearing loss of 30 dB HL or more at the age of 70 years, as do 55% of men and 45% of women at the age of 80 years (Roth et al., 2011). Although hearing worsens with age, the severity of the hearing problem at any given age varies greatly (Gates and Mills, 2005). Hearing loss affects an individual's psychosocial situation; untreated hearing impairment contributes, in fact, to social isolation, depression, and loss of self-esteem. Hearing impairment in advanced age has also been implicated as a possible cofactor in cognitive decline, dementia and psychiatric disorders (Lin et al., 2013; Quaranta et al., 2015; Gallacher et al., 2012). However, the directionality of this association is now being investigated further and has, as we shall see, have interested part of my research work.

I. 2. 1. Auditory perceptual disorders

-*Tinnitus*: Tinnitus is defined as an abnormal auditory sensation that appears without external physical stimuli (Jastreboff, 1990). It is a non-specific symptom related to almost all diseases of the auditory system. However, it can also be present in apparently hearing-impaired individuals. The sensation is generally of an elementary nature—descriptions of hissing, sizzling, and ringing are common, although, in some cases, more complex sounds such as voices or music are perceived. Tinnitus can be constant or intermittent, and many patients experience more than one sound. It can be localised to one or both ears or centrally within the head, although some patients describe an external point of origin. The onset of tinnitus can be abrupt, but it is insidious in most cases. The perceived intensity can vary; for some people, exacerbation alongside stress arousal is clear. The genesis of tinnitus is complex and unclear: it involves not only the peripheral auditory system but also the central auditory pathways and the areas of the brain devoted to processing attention and emotions (Ambrosetti et al., 2018). Chronic tinnitus can be accompanied by psychiatric diseases such as depression and anxiety (Langguth et al., 2011), insomnia (Asnis et al., 2018) and in extreme cases, intractable tinnitus may lead to suicide (Johnston and Walker, 1996). Moreover, it has been frequently noted that tinnitus is associated with reduced cognitive functions (Gatehouse, 1991) like working memory and attention (Rossiter et al., 2006).

The heterogeneity of tinnitus experience to which I have just referred is substantial and has hampered both basic science and treatment research (Baguley, 2013) posing important questions in the field of auditory neuroscience to which I have attempted to offer answers as part of my academic career.

-*Hyperacusis*: The term hyperacusis refers to abnormally strong reactions occurring within the auditory pathways resulting from exposure to moderate sound; consequently, patients express reduced tolerance to suprathreshold sounds (Jastreboff and Jastreboff, 2000). This phenomenon should not be confused with *recruitment*, an abnormally rapid growth of loudness in the vicinity of the elevated threshold (Fowler, 1973; Buus and Florentine, 2002). Hyperacusis has several potential mechanisms which are not mutually exclusive; as with tinnitus, the patient population is likely to be heterogeneous. It is prevalent in Williams syndrome, a disorder characterised by deficits in conceptual reasoning, problem-solving, motor control, arithmetic ability and spatial cognition (Levitin et al., 2003). Moreover, hyperacusis appears to increase in extent at times of tiredness, or stress (Sahley and Nodar, 2001) and may evoke anxiety and even fear. This can be true for specific sounds or sounds in general. The links between the central auditory system and areas of the brain implicated in anxiety and fear are now under scrutiny (Baguley, 2003).

-*Misophonia* as hyperacusis falls under sound tolerance conditions and refers to intense emotional reactions to certain sounds, regardless of the loudness of those sounds (Palumbo et al., 2018). Misophonic reactions resulting from trigger sounds involve arousal of the sympathetic nervous system and emotional distress (Erfanian et al., 2019).

-*Phonophobia* finally is defined as a persistent, abnormal, and unwarranted fear of sound. Often, these are normal environmental sounds (for example, traffic, kitchen sounds, doors closing, or even loud speech) that cannot, under any circumstances be damaging (Henry et al., 2022).

As I have illustrated so far, although hearing appears to most listeners to be a simple, innate and stainless ability, it is the result of a complex process involving various anatomical systems and functions whose essential importance is often only realised when disturbances accompany it. Confirming that hearing is not a trivial means of communicating with the world but an incredibly connected and fundamental multicomponent function in the different spheres of human life, interpersonal and intrapersonal.
In addition to hearing loss, tinnitus, and hyperacusis, a complex aspect to consider in the context of hearing is ‘listening effort’ precisely because of its multi-component characteristics.

I. 2. 2. Listening Effort

Until now, has been described the typical pathway that the sound wave takes from the air along the anatomical districts of the auditory system in order to stimulate the auditory nerve that transmits the acoustical signal to the brain. This mechanistic, one-way vision of hearing is now outdated because, although the sense of hearing is transmitted through a perfect chain, it is the whole brain that enables us to listen earing is perceiving sound, listening is making sense of sound (Kraus, 2021). And in this whole process involving perception, cognition and emotion, the cognitive load in listening is a crucial component. Listening effort (LE) is defined as a specific form of mental effort that occurs when a task involves listening (Pichora-Fuller et al., 2016). So, Pichora-Fueller's definition takes Kanheman's work on attention and effort and extends it to hearing. Kahneman, in his Capacity Model of Attention— which relies on the idea that the information processing capacity in humans is limited and that the ability to perform a task depends on the resources available and those required by the task— described attention as a reservoir of mental energy from which resources are drawn to meet situational attentional demands for task processing. He then argued that mental effort reflects variations in processing demands (Kahneman, 1973).

Reflecting, everyday listening frequently occurs in the context of acoustic challenges that degrade the auditory signal (Mattys et al., 2012). External sources of the acoustic challenge include background noise, competing speech, or talkers with a foreign accent (Van Engen and Peelle, 2014). Moreover, even when the external auditory signal is perfectly clear, hearing impairment reduces the fidelity of the information reaching a listener’s perceptual system (Peelle, 2014). Clearly, for the normal-hearing population, everyday listening is generally a relatively effortless process. In fact, when listening in noisy environments, the brain carries out all of the necessary ‘backstage operations’ that permit the selective processing of a particular sound and simultaneous filtering out of irrelevant information (McGarrigle et al., 2014). In contrast, for individuals with hearing loss, listening (aided or unaided) is often reported to be considerably taxing (Kramer et al., 2006). Measurements of listening effort arising from, respectively, the listener's difficulties, speech characteristics, and environment, encompass a wide range of techniques. These techniques can be categorised into three main subgroups (Shields et al., 2022) (figure 12).

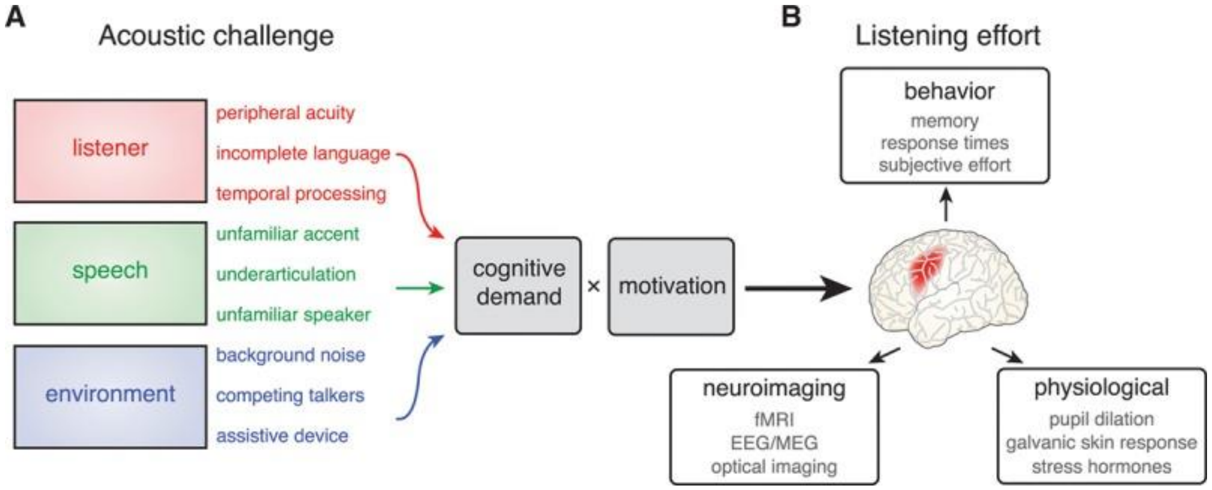


Figure 12. Mechanisms leading to listening effort and measures. (A) The overall acoustic challenge experienced by a given listener is a combination of individual hearing ability and external acoustic characteristics (including speech quality and background noise). (Note only a subset of these conditions are directly addressed in the main text.) Acoustic challenge increases cognitive demand, which is a key contributor to listening effort (moderated by motivation). When speech is not easily matched to a listener's expectation, additional neural processing is frequently required. (B), Increases in listening effort can be observed through functional brain imaging, are reflected in physiological responses outside the brain, and frequently result in measurable differences in behavior. (Source: Peelle, 2014).

(1) *Self-report measures*: come in the form of a closed-set questionnaire or rating scale, for example the multi-dimensional speech, spatial, and qualities (SSQ) hearing scale (Gatehouse and Noble, 2004) which measures the extent of listening difficulties experienced in various real-world settings. Self-report judgments of effort are also used during experimental listening tasks. For example, in one large-scale study, participants were asked to indicate (on a 100-point scale) how 'effortful' they found each particular trial in an auditory profile test battery (Van Esch et al., 2013). Self-report measures of LE are quick and easy to deliver and do not require particular expertise to administer and interpret. These measures provide an insight into how effortful speech processing is experienced by the individual. However, the limitations of self-report measures stem from their subjective nature and are so affected by individual differences in interpreting questionnaires (Alhanbali et al., 2019).

(2) *Cognitive and behavioural measures*. Behavioural responses in listening tasks have also been used to index listening effort. Such measures can be categorised into single-task and multi-tasking paradigms. In the first, participants respond to stimuli either by means of verbally identifying the heard word/sentence (Gatehouse and Gordon, 1990) by pressing a response button (Houben et al., 2013). The second type of task builds on Kahneman's model introduced earlier, according to which there is only so much cognitive resource (i.e. 'energy') that needs to be efficiently distributed among the various mental operations. Thus, when performing two tasks simultaneously, if one (e.g. the primary task) becomes more demanding, this will result in a decrease in performance in the secondary task. (secondary) task. Performance on the latter can thus be interpreted as a reflection of the amount of effort allocated to the

primary task (Cennamo, 1992; Karatekin et al., 2004). The multi-tasking method appears to have good face validity with regard to speech processing in realistic environments (McGarrigle et al., 2014)
While listening effort, as brilliantly noted by Pichora-Fuller and colleagues (Pichora-Fuller et al., 2016) has sometimes been described as a form of difficulty that occurs in tasks involving listening, in reality, listening involves both auditory and cognitive processing, as suggested by the definition of listening as 'listening with intention and attention' (Kiessling et al., 2003). In fact, following the insight that auditory and cognitive functions are interdependent during the comprehension of spoken language and in many other listening tasks, see (Cherry, 1953) for an early example, the field of cognitive hearing science developed over the past decades. Furthermore, there is a recognised approach to the topic of listening effort in the literature that applies cognitive theories of attention, working memory and speed of processing to the cognitive factors involved in listening, speech comprehension and ageing (cf. Cohen, 1987; Craik and Salthouse, 2011). Therefore, it can be deduced how the study of the cognitive listening component can offer more insight into LE and its effects on both the physical and cognitive levels.
Indeed, in the long term, if listening in everyday activities often requires more effort than listeners are able or willing to expend, they may develop chronic stress and withdraw from social interaction, with negative consequences on cognition, general health, well-being and quality of life; (Pichora-Fuller, 2016). Moreover, LE is a valuable and essential notion to measure because it is among the primary complaints of people with hearing loss (Winn and Teece, 2021).

(3) *Physiological measures* refer to the recording of changes in central and/or autonomic nervous system activity during task performance providing a way of examining how biology influences communication, and, in turn, communication influences biological changes within the body (Priem, 2015; McGarrigle et al., 2014). Moreover, psychophysiological measures, assess the interaction between psychological and physical states using a variety of instruments in both laboratory and naturalistic settings (Gaffey and Wirth, 2014), indeed, psychophysiology relates individual mental functions to physiological signatures, as exemplified in recent years by functional brain imaging (Gray et al., 2009).
Listening effort-related changes in central nervous system activity have been investigated in normal and hearing-impaired subjects, using functional magnetic resonance imaging (fMRI) (Wild et al., 2012); (Dimitrijevic et al., 2019); electroencephalography (EEG) (Cartocci et al., 2018); (Wisniewski et al., 2015), and event-related potentials (ERPs) (Bertoli and Bodmer, 2014). Autonomic nervous system activity has also been examined for evidence of listening effort-related changes, including changes in skin conductance (Mackersie and Cones, 2011) and pupil dilation (Zekveld et al., 2010; Neagu et al., 2023). Such experiments typically involve a behavioural task with several conditions which vary in degree of difficulty. Assuming comparable behavioural performance between conditions, systematic physiological changes that occur in the more-challenging condition/s are generally attributed to listening effort (Shields et al., 2022). As will be seen in the part III of this thesis, I conducted specific studies to

answer two scientific questions (C, D) analysing the electroencephalographic characteristics of listening under different noise conditions.

I. 2. 3. Diagnostic Tests

Hearing tests of the peripheral auditory system are well established, and the pure tone audiometric (PTA) examination is the gold standard test of audiological examination to assess auditory ‘acuity’ that is, how sensitive the auditory system is to sound; auditory acuity is assessed by determining the intensity at which a tone is just audible (McCullagh, 2013). It allows the distinction between conductive, i.e., outer- and middle-ear, and sensorineural, i.e., cochlear hearing loss, and to describe the configuration of the hearing thresholds in terms of severity and the frequency affected (Davies, 2016). PTA is a subjective, behavioural measurement of a hearing threshold, as it relies on patient responses to pure-tone stimuli (Prosser and Martini, 2013). Pure-tone evaluations are performed in sound-treated rooms to eliminate ambient noise while obtaining hearing thresholds for both air-conducted and bone-conducted sounds. *Air conduction* tones are presented through inserted earphones or over-the-ear headphones. *Bone conduction* thresholds are obtained via an oscillating transducer placed on the mastoid process. Providing ear specific thresholds and uses frequency-specific pure tones to give place-specific responses, so that the configuration of a hearing loss can be identified. This method (Roeser, 2013) is used to measure the threshold of hearing perception for pure tones in the range of frequencies across the speech spectrum (500 to 4000 Hz) to 8000 Hz at the upper limits of normal hearing (25 to 30 dB for adults, and 15 to 20 dB for children), in octave intervals (nb one octave= doubling of frequency). As the PTA increases, the hearing ability decreases. Normal hearing for speech is observed in adults with PTAs of 25 dBHL or less. At a PTA of around 40 dBHL in both ears, most people are considered functionally impaired and could benefit from amplification. Severe to profound losses occur when PTAs are greater than 70 dBHL. At this level, hearing aids provide limited benefits, and cochlear implants may be considered (Gates and Hoffman, 2007). Practically, the in-cabin test is performed by means of an audiometer (acoustic signal generator) calibrated in decibel hearing level (dBHL) according to the International Organization for Standardization (ISO) norm (ISO 11957:1996, 1996)[6] (ISO, 2023). Briefly, the patient is asked to indicate when a given pure tone, sent at gradually decreasing intensity in the headphones for each side, is no longer heard (descending method) or when, by progressively increasing the intensity of the sound, the subject continues to hear it (ascending method). The responses are marked on a graph (audiogram) using different symbols and colours according to the ear being analysed. In the case of unilateral or asymmetrical hearing loss, the clinician performs “masking”. This term is used to indicate the functional exclusion of the ear contralateral to the one whose threshold is to

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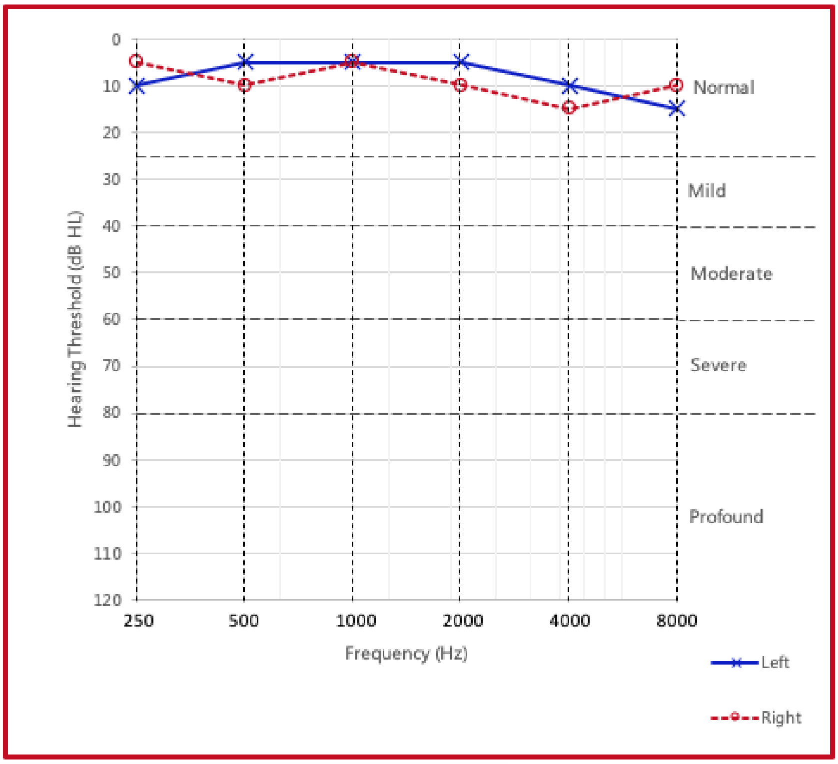
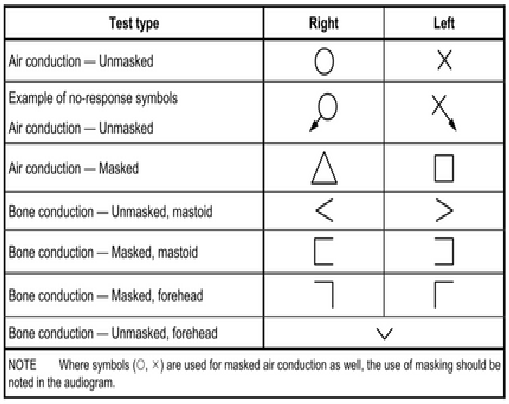
[6] https://www.iso.org/obp/ui/#iso:std:iso:11957:ed-1:v1:e

be measured. Depending on whether a hearing- or hearing-impaired subject is examined, various curves will be outlined on the audiogram (Cianfrone, 1986), (figure 13).





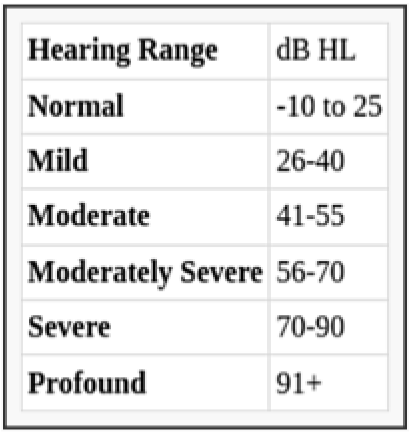
Test type	Right	Left
Air conduction — Unmasked	○	×
Example of no-response symbols Air conduction — Unmasked	○↙	×↘
Air conduction — Masked	△	□
Bone conduction — Unmasked, mastoid	<	>
Bone conduction — Masked, mastoid	[	]
Bone conduction — Masked, forehead	┐	┌
Bone conduction — Unmasked, forehead	∨	

NOTE Where symbols (○, ×) are used for masked air conduction as well, the use of masking should be noted in the audiogram.

Figure 13. The graphic represents an audiogram that plots hearing thresholds across different standardized frequencies as measured by an audiometer. The horizontal axis (highlighted below) is the frequencies tested during the hearing test, expressed in terms of Hertz (Hz), going from left to right, the frequency increases. The vertical axis (highlighted below) is the hearing thresholds measured during the hearing test, expressed in decibel hearing level (dB HL). The numbers (0,10,..120) indicate the volume or loudness of the sounds required for the subject to hear. The higher the number, the louder the sound. Hearing sound from a smaller number represents a better hearing result. Concerning the symbols, according to the legend for this audiogram, the left ear thresholds are marked with blue X’s, and the right ear thresholds are marked with red O’s. (Source: hkincus.com, 2023).

The table under the audiogram is an audiogram symbols key in the format specified by "ISO 8253-1:2010 Acoustics — Audiometric test methods " (ISO 8253-1:2010 Acoustic).

Generally, a person with a threshold within 15 dB HL is considered clinically normal hearing (Prosser, 2018). The figure 14 shows a functional classification.



Hearing Range	dB HL
Normal	-10 to 25
Mild	26-40
Moderate	41-55
Moderately Severe	56-70
Severe	70-90
Profound	91+

Figure 14. Classification of hearing loss. (Source: Clark, 1981).

However, it is not always possible to obtain reliable hearing information for infants and children using pure-tone audiometry due to a lack of cooperation and an inability to understand the test. As an objective alternative method for acquiring pure tone audiograms for infants and children not suited to behaviour observation audiometry, the electrophysiologic testing with auditory potentials, e.g. the auditory brain stem response (ABR), can be used to identify sites of lesions in the eight nerve, brainstem, and more centrally (Sininger et al., 2000). This technique can, however, face difficulties in the determination of the participating frequency ranges because the test collects responses from the whole basement membrane with a stimulus tone of short duration. As a result, the ABR method has little frequency specificity (Ahn et al., 2007).

Moreover, although PTA has many clinical benefits, it is not perfect at identifying all losses, such as ‘dead regions’ of the cochlea and neuropathies such as central auditory processing disorder (CAPD) (Cacace and McFarland, 1998). For these diseases, the most sensitive and specific diagnostic tool is indeed the ABR (Hosford-Dunn, 1985). With advancing age, individuals present a sloping hearing loss in the high frequencies, which may be due to normal senescence of the hearing apparatus (the previously described presbycusis) or to the accumulation of specific damaging factors over time (Gordon-Salant et al., 2010). Even though the loss of hearing perception in an adult can lead to major disturbances and impact well-being even in globally healthy people, it is conceivable how severe hearing loss at birth can have an unfavourable influence with catastrophic consequences on a child’s psychological, linguistic and social development. Fortunately, medical research has made it possible, as it will be shown shortly, to cure the once irreparable deafness, allowing the transmission of the acoustic wave from the world to the cerebral cortexes of children (and, also in adults), thus supporting them in healthy development.

I. 3. The Cochlear Implant

Hearing research is one of the great scientific success stories of the last 100 years. Not only so many exciting discoveries have been made about the nature of sound and the workings of the ear and the auditory brain, but these discoveries have also driven many immensely useful advantageous technological developments in entrainment and communications as well as for clinical applications designs to help patients with hearing impairment (Schnupp et al., 2011). A concrete representative of this hearing synergy is the cochlear implant. The cochlear implant (CI), otherwise known as a cochlear prosthesis, is a true artificial sense organ. In fact, it can directly stimulate, through electrical signals, the remaining fibres of the auditory nerve, generating auditory sensations in the cortical areas of the central nervous system in individuals with total or profound deafness. The cochlear implant, just like a bionic ear, converts acoustic signals into electrical impulses that, bypassing the damaged structures of the inner ear, directly stimulate the auditory nerve (Guida et al., 2018).

CIs are de facto, the only medical intervention that can restore partial hearing to a totally deafened person via electric stimulation of the residual auditory nerve (Zeng, 2004). The development of cochlear implants can be traced back at least 200 years to the brilliant Italian scientist Alessandro Volta (1745-1827). He invented the battery and used it as a research tool to demonstrate that electric stimulation could directly evoke auditory, visual, olfactory, and touch sensations in humans (Volta, 1800).

The Italian scientist reported perceiving auditory sensations after applying the two electrodes of the battery he had invented in the external auditory canal. Volta thus wrote to the president of the Royal Society.

"*. . . at the moment when the circuit was completed, I received a shock in the head, and some moments after I began to hear a sound, or rather noise in the ears, which I cannot well define: it was a kind of crackling with shocks, as if some paste or tenacious matter had been boiling . . . The disagreeable sensation, which I believe might be dangerous because of the shock in the brain, prevented me from repeating this experiment . . .*" (Volta, 1800, p. 427).

However, we have to wait until the 20th century to talk about cochlear implants, resulting from an inexhaustible synergy between research and medical clinics. In fact, electrophysiologist Djourno (1904-1996) and otolaryngologist Eyriès (1908-1996)'s work, dating to 1957, definitively spurred the development of the CI. Strictly speaking, the history of CIs began in the early 1960s with the experiments that William House conducted in collaboration with the Massachusetts Institute of Technology's scientists. However, it took more than ten years before the application of CI on a cohort of patients was realised (Mudry and Mills, 2013).

By the end of the 1980s, CI became the predominant treatment for profound deafness in the United States, Europe, and Australia, bringing about a new controversy over the "origins" of the technology, as well as controversy about its application among the Deaf community (Blume, 2010).

The genesis of the cochlear implant was briefly mentioned to show how its development has resulted from human scientific imagination combined with clinical practice from the beginning. This synergy continues to this day, and part of the present work is intended to build on it.

Clinically, CI has represented, for almost 25 years now, the gold standard in the treatment of children born deaf and for postlingually deafened adults (Stöver and Lenarz, 2009) and is constantly being improved and adapted to patients' actual needs (Blebea et al., 2022). Furthermore, taking advantage of explosive growth and innovations in technology, particularly in microelectronics in the last two decades, cochlear implants have evolved from analogical to digital, from a single electrode to multiple electrodes, from percutaneous plugs to transcutaneous transmission, and from simple modulation to complicated feature extraction processing (Zeng, 2004).

Moreover, many different designs of cochlear implants have been described, including both commercial and experimental systems, but all designs have general features in common. I will limit myself to describing its main characteristics, postponing the technical details to other works (i.e DeSaSouza, 2022) since it is not the central focus of my research. Modern CIs consist, indeed of two principal components (Clark, 2003; Guida et al., 2018; Marsella and Scorpecci, 2018):

- one external, consisting of a processor (speech processor) control unit and transmission unit comprising an external antenna and magnet;

- an internal one consisting of a receiver unit (internal antenna and magnet), stimulation unit (receiver-stimulator) and an electrode array. (Figure 15).

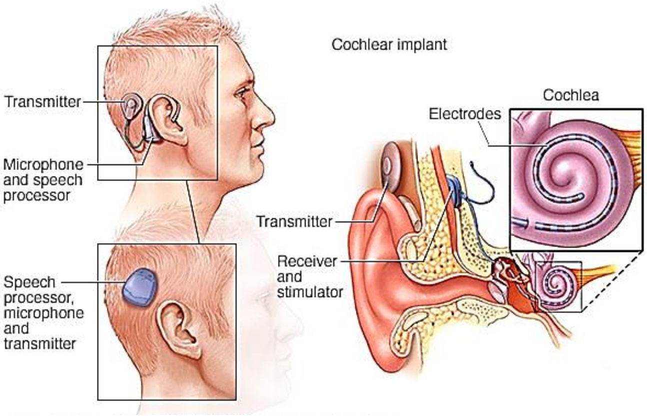


Figure 15. How cochlear implants work. A cochlear implant uses a sound processor worn behind the ear. A transmitter sends sound signals to a receiver and stimulator implanted under the skin. They stimulate the auditory nerve with electrodes threaded into the cochlea. Some types of cochlear implants have one outside unit with a speech processor, microphone and transmitter combined (lower left). Others have these as separate outside parts (upper left and on the right). (Source: Mayoclinic, s.d.).

The speech/sound processor (figure 16), worn behind the pinna, decodes sounds picked up from the external environment via a microphone and low-frequency circuits, transmits them to the internal component (receiver-stimulator) in the form of electrical stimuli. It consists of a microphone analogue-to-digital converter (ADC), signal pre-processing unit, data filter unit, signal amplitude mapping unit and radio frequency converter.

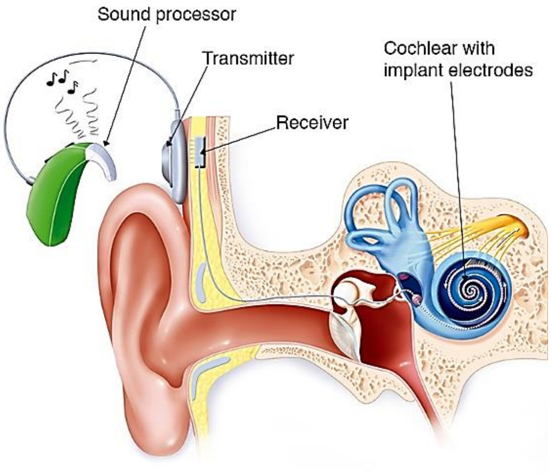


Figure 16. Synthetic image of a cochlear implant. The external sound processor converts sound into a sequence of electrical impulses and sends these signals *via* a transmitter coil to the internal receiver stimulator. The internal device, located under the skin in a cavity created in the skull bone, processes the electrical signal and transmits them *via* electrodes to the spiral ganglion cells of the cochlear nerve. Both internal and external devices are equipped with magnets. (Source: Huber, 2022).

The pre-processing unit thus filters the signal and can act on the microphone characteristics by modifying its gain and sensitivity. Once pre-processed, the electrical signal is analysed by a band-pass filter capable of breaking the signal down into frequency bands and digitised, thus allowing the signal to be converted from analogue to digital.

The receiver–stimulator package is placed subcutaneously and is fixed on the mastoid bone; this package receives electromagnetic signals and delivers them through lead wires to an electrode array placed in the cochlea. The electrode contacts can exploit the tonotopic arrangement of nerve fibres, with high sound frequencies represented at the cochlear base and low frequencies at the apex. Activation of the implant generates an electrical response in selective auditory nerve fibres, which is carried to the auditory cortex and is interpreted as auditory input.

Electrode stimulation can take place in three different ways (figure 17). The three main configurations available with existing devices are known as monopolar, bipolar, and common ground. The *monopolar* configuration comprises an active electrode located in or close to the cochlea, and one or more separate electrodes located further away. In multiple-electrode implants employing monopolar stimulation, the active electrodes must be located close to the neural population so that, ideally, stimulation on each electrode excites a spatially distinct set of neurons and consequently elicits a perceptually discriminable auditory sensation. In the *bipolar* configuration, currents are passed between two electrodes, which are located relatively close to the auditory neurons. Finally, the *common-ground* mode, one active electrode is selected, and many or all of the remaining intracochlear electrodes are used together as the return path for the stimulating current (McDermott, 2004).

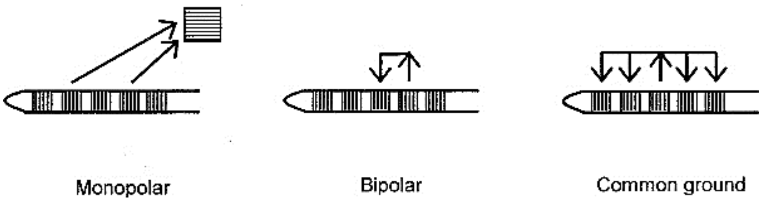


Figure 17. Three types of electrode configurations used in multiple-channel implants. The illustration at the left shows the *monopolar mode*, in which current from the active electrodes on the intracochlear array flows to a single "ground" electrode located remotely. The centre illustration shows the *bipolar mode*, in which current passes between two active electrodes located nearby on the array. Finally, the illustration at the right shows the *common ground* mode, in which current from one intracochlear electrode flows to most or all of the remaining electrodes on the array. (Source: McDermott, 2004).

I. 3.1. Indications for cochlear implantations in Children and Adults

Although cochlear implantation has been an approved method of treating profound, bilateral, sensorineural hearing loss for persons since the mid-1980s (House and Berliner, 1991), there is a continuous, gradual and significant widening of the indications for this procedure (Szyfter et al., 2019; Deep et al., 2019; ASHA, 2004; Quaranta et al., 2009). They concern the age of eligible patients, implantation in patients with partially preserved hearing, as well as treatment for patients with difficult anatomical conditions. In addition, in many countries bilateral implantations are commonly performed, and more centres recommend this treatment in the case of unilateral deafness or asymmetric hearing loss, especially with the accompanying tinnitus in the deaf ear.

Children: The indications for cochlear implantation in children are constantly evolving and conditioned by technological developments, knowledge of the disease, the experience of the doctors involved and the states. According to Italian national guidelines, cochlear implantation is recommended in children with severe-profound bilateral sensorineural hearing loss and defined as a threshold >80 dB for frequencies 0.5-2 kHz (Quaranta et al., 2015). While for the UK, CI is recommended in children with bilateral profound sensorineural hearing loss (>90 HL) for frequencies 2-4 kHz (National Institute for Health and Care Excellence, 2019). While the French guidelines indicate implantation in children in cases of profound deafness if the prothesis does not allow speech development and in cases of profound deafness based on the score obtained in speech audiometry tests carried out at 60 dB (de Santé HA, 2007).

However, beyond the insignificant threshold differences, all guidelines agree on the need, before indicating cochlear implantation, to assess for at least three months the lack of benefit of hearing aid fitting defined as failure to achieve age-appropriate perception, comprehension and production skills, developmental stage and cognitive abilities (Marsella and Scorpecci, 2018).

The timing of surgery in children is still a ground of dispute between states. While, for example, the Italian guidelines provide for prothesis even at the age of less than 12 months in particular complex diagnostic situations (Quaranta et al., 2009), in the United States the Food and Drug Administration requires one to wait until the child is one year old, dropping from 2020 to 9 months old only for particular processors (US Food and Drug Administration, 2020) while the British guidelines do not set a lower age limit (National Institute for Health and Care Excellence , 2019).

However, it is scientifically documented that the fundamental period for learning language is the first 5-6 years of life (Cosetti and Roland, 2010; Taylor, 2007; Piaget, 1967) and that hearing deprivation before and during this period produces irreversible damage to learning itself (Lederberg et al., 2013). This is proven by neurophysiological and cognitive scientific evidence showing that there are *sensitive periods* —which will soon be further explored— for developing language capacity and clearly of all the socio-communicative, cognitive, and emotional skills that come with it. It goes without saying that speech quality, language skills, expressive and receptive vocabulary are improved by exposure to aural language from as early as possible (Kileny et al., 2001). Therefore, early implantation facilitates improved development of speech perception skills in profoundly deaf children (Zwolan et al., 2004).

Adolescents: The indication of cochlear implantation in preverbal deaf adolescents is extremely delicate and difficult to frame, as brain plasticity and auditory memory at this age of life are poorly utilised (Guida et al., 2018). The results show that speech perception scores after implantation almost universally improve but are worse when compared with age-matched control peers undergoing implantation at an earlier age (Santarelli et al., 2008; Shpak et al., 2009).

Adults: In postverbal deaf adults, in cases of bilateral, profound or severe recent onset deafness whose verbal discrimination is zero or less than 30% regardless of hearing loss. Motivation must be high but not excessive, lip-reading present, and the neuropsychological picture normal (Guida et al., 2018). Finally, adults with different lateralisation of deafness (i.e. asymmetric hearing loss, single side deafness) reporting severe tinnitus in the deaf ears, which cannot be treated with other hearing aids, can benefit from cochlear implantation. Clearly these interventions must be considered on a case-by-case basis (Olze et al., 2022).

I. 4. Neural Plasticity

Early experiences are able to shape human mental capacities ranging from the awareness of making sense of what we see to the ability to have appropriate social interactions (Sanes and Jessell, 2019). The first evidence of how social and perceptual experience is fundamental to human development was obtained from observational research by Rene Spitz (1887-1974) in the early 1930s (Spitz, 1989). The psychoanalyst compared the development of children reared in orphanage with that of children reared in a women's prison kindergarten (Spitz, 1945). The children in the prison kindergarten saw their mothers showering them affectionately in moments of encounter. Whereas, the children in the orphanage, being cared for by nannies, had less contact with the caregivers. In addition, the cots where the children slept differed: in the prison kindergarten, they were open, and the children could observe the activity around them, while, in the orphanage, the bars of the cots were covered with sheets which prevented the children from looking out. As a result, the children in the orphanage lived in sensory and social deprivation conditions. Observation during the first years of the life of the infants born in the two situations showed that growing up in the orphanage resulted in the development of motor, intellectual and emotional deficits while the children born in the prison annexe kindergarten did not develop such deficits.

Clearly, no objective biological data can be derived from this observational study, and no significant neurophysiological conclusions can be drawn. Spitzôs observations, however, have drawn attention to what *sensory deprivation* entails for human development, confirming the fundamental importance of experience on brain function and the ability of experience to shape perception corroborating and suggesting that one of the most essential functions of the nervous system is to obtain information about events in the external environment (King, 1999) and this is made possible by brain plasticity. A recent event of ñglobal sensory deprivationò can certainly be considered the quarantine period due to the covid 19 pandemic. In this context, as will be seen in the experimental part of this work, I had the idea of investigating how such macro sensory deprivation was affecting the well-being of students with and without hearing impairment (see work H and I in the experimental research section).

The first use of the term 'plasticity' to describe the nervous system can be traced back to experimental psychologist William James (1842-1910) more than a century ago. In his classic textbook Principles of Psychology (James, 1890), James stated: *"Plasticity, then, in the wide sense of the word, means the possession of a structure weak enough to yield to an influence, but strong enough not to yield all at once. Organic matter, especially nervous tissue, seems endowed with a very extraordinary degree of plasticity".*

James borrowed the term from physics and materials science (for details in physics see Lubliner, 2008), where it was used to describe the ability of certain physical materials to alter their shape in response to external forces or internal tensions (Warraich and Kleim, 2010). With sufficient force and repetition, a piece of metal can be bent or modelled into a different shape. James proposed that similar changes could occur in the nervous system in response to external forces, such as behavioural training, or internal stresses, such as disease or injury. He referred to the brain as "*an organ in which currents pouring in from the sense organs make with extreme facility paths which do not easily disappear"* (James, 1890) And, it is possible to add here, paths traced by waves.

During the same period, the pioneering work of several neurobiologists, including Ramon Y. Cajal (1852-1934) and Charles Sherrington (1857-1942), demonstrated that the nervous system was indeed composed of individual units (neurons) connected by specialised junctions (synapses) (Cajal, 1889). Neurobiological evidence now existed for the pathways in the nervous system that James proposed were capable of plasticity (King, 1999). Furthermore, as seen previously, since the sensory experience of the outside world is transformed in the brain in the form of electrical activity, it is conceivable that the type of electrical signals that arise in neural circuits may alter the circuits themselves.

Focusing his scientific attention from an internal perspective on neuronal mechanisms, compared to James' insights, Donald Hebb (1904-1958)'s theory of cognition and learning proposed that organised activity within a brain circuit could be sustained and, significantly, re-implemented in the future, if neurons were organised so that a circuit could be self-exciting and therefore sustain its activity for a while and its connections between neurons were selectively strengthened between co-active neurons. This idea is often summarised by the phrase 'neurons that fire together wire together' (Hebb, 1949). Hebb's ideas predated the evidence for 'Hebbian plasticity' by decades, but his theories now have extensive empirical support and underpin our current understanding of development, learning, and memory (Power and Schlaggar, 2017). Numerous studies and in-depth studies followed Hebb's ideas, clearly not being the subject of the present work; they will not be examined in detail for which reference is made to other sources, for example (Sweatt, 2016).

However, these slight hints, like buoys placed along the course followed by this work, indirectly underline the importance of neuronal plasticity for developing sensory systems. Specifically, the plasticity of the auditory nervous system is vital throughout life, but especially in the early years, as we will shortly explore. Indeed, this physiological feature plays a crucial role in developing skills in infancy and early life that allow exposure to meaningful sounds to form the neural basis of understanding. (Møller, 2018a). In practice, as Kraus summarises, "the sounds of our lives shape our brain" (Kraus, 2021) In fact, once the ear becomes capable of transducing sounds and the central nervous system is capable of encoding information with adequate precision, the stimulus-evoked activity becomes available for making further refinements in the neural circuitry underlying the auditory space map. As with other systems, the contribution of experience to auditory map development has been assessed by observing the physiologic consequences of raising animals with altered sensory inputs (King, 1999). It is, in fact precisely by observing sensory deprivation that it was realised that sensory stimulation is essential to lay the foundations for healthy physical, psychological and cognitive development.

Delving into, the suffix "de" in front of the word "deprivation" indicates the absence of what is necessary and thus emphasises that auditory information is considered fundamental to the human being (Di

Bernardino, 2019). If the hearing is impaired or absent, the individual is not only deprived of a specific detail: a complex situation is configured that depends on the extent, the cause and, above all, the temporal moment in which the deprivation manifests itself. In fact, there are time windows of early postnatal development when structural and functional neural circuits are shaped by passive experience with the external environment (Persic et al., 2020). The first experimental studies on this are due to David Hubel (1926-2013) and Torsten Wiesel (1924), who investigated the effect of visual sensory deprivation in an animal model. Without dwelling further on the studies conducted by Hubel and Wiesel, not within the domain of the present work, their ground breaking research scientifically demonstrated that early experience is a critical and obligatory factor for the cortex (visual in this case) to develop its standard typical structure and function, opening up research into critical periods of development (Hubel and Wiesel, 1970). Brain development, in fact, includes periods of higher susceptibility to alterations by an experience called *sensitive periods* (or *critical periods* in terms of behaviour on the basis that the absence of specific juvenile experiences cannot be fully compensated later in life) (Kral, 2013) (figure 18). And, early in development, the existence of critical periods for experience-dependent plasticity has been demonstrated for the visual, auditory and somatosensory systems (Berardi et al., 2000) and in fundamental abilities like language learning in humans (Friedmann and Rusou, 2015).

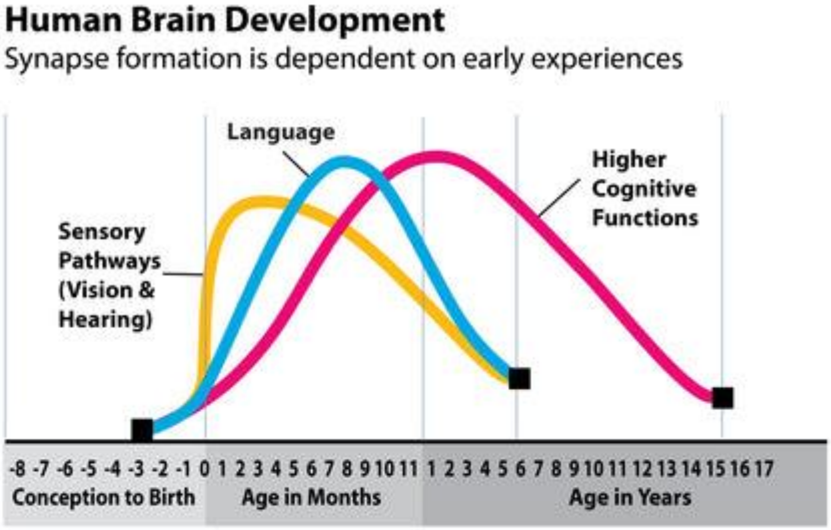


Figure 18. Human brain development by age. Sensory pathways peak in the first few months of life, whereas higher cognitive function does not develop until much later. Higher cognitive function includes attention, planning, problem-solving, and decision-making. The timing is genetic, but early experiences determine whether the circuits are strong or weak. (Source: Nelson, 2000).

In fact, during learning, a sensory stimulus gains new behavioural significance, resulting in a dynamic reorganisation of the representation of the features and objects associated with that sensory stimulus

(Gilbert and Sigman, 2007). In the auditory system, the most extensively studied critical period is that for the calibration of the auditory space map by visual input (King and Moore, 1991). Moreover, some auditory functions show optimal performance if learned early in life: e.g. musical experience has the most pronounced effects on performance during early childhood (Penhune, 2011). Interestingly, musical training in infancy leads to an expanded auditory cortical representation, but only if practising began before the age of 9 (Pantev et al., 1998).Also, language learning is easiest early in life. In fact, the best-known auditory examples of critical periods were observed in language development (Kral, 2013). The first theory regarding the critical period for language was suggested by Penfield and Roberts (Penfield and Roberts, 1959) and later developed by Lenneberg and Chomsky (Lenneberg, 1967; Chomsky, 1976). Various hypotheses on the temporality of the critical window for language acquisition have ignited scientific debate. For example, while for Lenneberg, the critical period for acquisition was from the age of two to puberty, for other researchers, it is possible to identify signs of language learning as early as the first days of life and even in utero (Dehaene-Lambertz et al., 2008; Mehler et al., 1988). Finally, it should be noted that some aspects of cortical organisation are also modifiable, by experience, in the adult. This is the case for the frequency map in the auditory cortex (Rauschecker, 1999).

In light of the above, as Friedmann and Rusou note (Friedmann and Rusou, 2015) it emerges that language input is fundamental to typical language acquisition and that the crucial role of this input can be studied well in populations of children born with auditory impairment. In fact, the duration of auditory deprivation, the age and developmental stage at which it occurs, and the cause and type of deafness are fundamental characteristics for understanding the multiple changes in the auditory pathways that occur at all levels: nerve, brainstem, cerebral cortex (Gordon et al., 2011). Researchers have provided evidence showing delayed cortical maturation without auditory stimulation, and additionally, that cochlear implants can restart this maturation process in children who received cochlear implants before about seven years old (Ponton et al., 1996a; Ponton et al., 1996b; Sharma et al., 2002). Correspondingly, language development measures have shown that children with CIs performed significantly better than expected from unimplanted deaf children and approached or reached the rate of language development observed in children with normal hearing (Moog and Geers, 1999; Svirsky et al., 2000).

More specifically, in bilaterally deaf children, CI has proven successful when implantation occurs within a sensitive period for central auditory development, wherein the auditory pathways are maximally plastic (Kral and Sharma, 2012). Finally, electroencephalographic studies carried out by Sharma and colleagues widely support that the optimal time to implant a young deaf child is within a sensitive period of ~ age 3.5 years in childhood (best by the first two years of life). After the overall sensitive period ends (~ age 7 years), there is a high likelihood of de-coupling of primary cortical areas from surrounding higher-order cortex and subsequent cross-modal recruitment of the higher-order auditory cortical areas by other modalities such as vision and somatosensorial. Thus, implantation within the sensitive period allows for normal auditory cortical maturation, providing ample opportunity for the appropriate acquisition of speech and oral language (Sharma and Campbell, 2011). In the case of prolonged sensory deprivation, a fundamental principle of neuroplasticity is that central sensory pathways undergo reorganisation (Campbell and Sharma , 2014). There is, indeed, ample scientific evidence of cross-modal reorganisation of the cerebral cortex in both blind (Kujala et al., 2000; Sadato et al., 2002) and deaf (Finney et al., 2001; Gilley et al., 2008; Sharma and Mitchell, 2013; Stropahl et al., 2017) experimental sample furthermore, for comprehensive review about plastic changes in the central auditory system see (Syka, 2002).

The non-exhaustive scientific evidence cited so far suggests the importance of investigating cross-modality and a-modality in children with cochlear implants, especially in certain cognitive and emotional abilities through specific tasks. This is to increase knowledge of how the absence of auditory stimulation in the months (or years) prior to cochlear implantation, may have influenced the organisation of cognitive functions, —as will be described below and investigated directly by me experimentally— essential for healthy development.

b. COGNITIVE AND EMOTIONAL WAVES

Until now, it has been investigated how the acoustic wave is transformed by the cochlea into an electrical wave capable of stimulating the auditory nerve to transmit the information to the higher areas up to the sensory cortices of the brain. However, stimulation of the sensory cortices (visual, auditory) is not sufficient for the behavioural responses related to stimulation to occur. For example, paying attention to a sound, listening to a speech, and keeping a telephone number in mind. For such observable behavioural responses to occur, that manifest the acquisition of so-called cognition or knowledge of the world, the sensory stimulus arriving in the sensory cortices must be processed by cortical association areas, the associative cortices. Indeed, the execution of behaviours depends on the integration of activity at various levels of the central nervous system. Nevertheless, different nervous system regions have been shown to participate in specific functions (Pandya and Yeterian, 1985).

Cortical association areas occupy most of the surface area of the brain cortex (figure 19) and are primarily responsible for the complex processing that occurs between the arrival of information at the primary sensory cortices and the generation of behavioural responses.

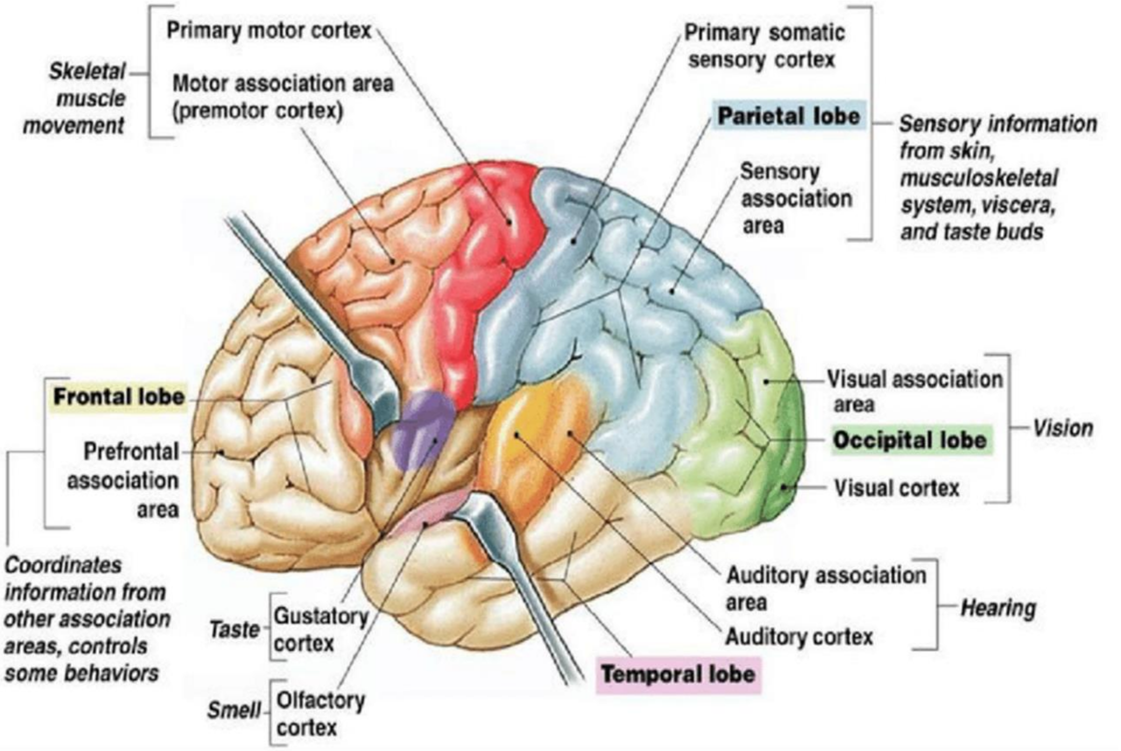


Figure 19. The human brain cortex and its areas. (Source: Nacy et al., 2016).

The different functions of the association areas of the cerebral cortex are known as “cognitive functions”, and together they enable us to acquire knowledge about the world (Purves et al., 2013a).

Cognitive function is hence, as will be discussed in more detail below, a broad term that refers to mental processes involved in the acquisition of knowledge, manipulation of information, and reasoning. including the domains of perception, memory, learning, attention, decision-making, and language abilities (Kiely, 2014).

What emerges is the complex role of interchange, reorganisation, and modulation played by the associative cortices, which retrieve information from multiple sources and, through their functioning, influence a wide range of cortical and subcortical targets. As if they were, harbour nodes where the different waves and currents find space and exchange modalities to depart, transformed, for further behavioural paths, to remain within the framework of the marine metaphor. Therefore, the afferences to the cortical association areas, include projections to the primary and secondary sensorial and motor cortices from the thalamus and brainstem. At the same time, they project to the hippocampus, thalamus, basal ganglia, cerebellum and other paths. Moreover, circuits are involved in transmitting sensory inputs into the amygdala, between amygdaloid subregions, and to efferent targets in cortical and subcortical regions, for specific emotional learning and memory processes (LeDoux, 1992).

It goes without saying how the study of cortical activations can bring further knowledge about cognitive functioning and, thus behaviour in subjects with and without sensory deficits. These aspects, which have opened diverse research questions in me, will be discussed later. For the moment, I will focus on punctually defining what cognitive functions are.

According to what was mentioned earlier, cognitive functions are mental abilities that allow the correct interpretation and management of environmental information (Forte et al., 2019). Cognitive functioning comprises multiple cognitive domains, such as memory, language, visuo-construction, perception, attention, and executive functions (Lezak et al., 2004). Furthermore, these skills are distributed along a continuum involving optimal cognitive functioning at one extreme and dementia at the other (Petersen, 2004).

I. 5. Working Memory

Working Memory (WM) refers to our ability to keep a small amount of information readily available for our current activities to support decisions, guide actions, make statements, keep track of conversations, navigate and support creative thinking and problem-solving, to remember to do things and to update what is going on around us throughout the day. In other words, it is a cognitive ability we use every waking moment (Logie et al., 2020b). The philosopher John Locke (1632-1704) was the first to identify the general concept behind WM, referring to the *contemplation* of ideas as opposed to the *storehouse* of ideas (Locke, 1860). The use of the term WM referring to this specific mental ability was introduced by Miller and colleagues (1960). However, it is the work of Alan Baddeley and colleagues that has made numerous theoretical and empirical contributions to WM's construct since the 1970s: but they are not the only ones! Over the past five decades, in fact, interest in this essential cognitive function has increased by delving deeper into its actual capacity (Logie, 2011), how it is developed and organised (Cowan, 2022), how its functions are implemented in brain structure, functioning (Smith and Jonides, 1997; Collette and Van der Linden, 2002), its cellular bases (Goldman-Rakic, 1995; Kamiński and Rutishauser, 2020) and how it is affected by structural brain damage (Müller and Knight, 2006; Ferber et al., 2020; Kimberg and Farah, 1993) and psychological (Alloway and Archibald, 2008; Alderson et al., 2013) and sensory disorders (Rönnberg, 2003; Gudi-Mindermann et al., 2018; Kronenberger et al., 2011; Heled et al., 2022).

But, apart from defining it, how can working memory be represented? A practical specification of the concept of working memory is due to Baddeley and Hitch (1974). In the original tripartite model of the researchers (figure 20), as an alternative to the short-term store model of Atkinson and Shiffrin (1968), WM would consist of an attentional system (the *central executive*), a flexible system responsible for the control and regulation of cognitive processes, that supervises and coordinates the two subsidiary systems (*slave systems*): the articulatory or *phonological loop*, responsible for the processing of linguistic information, and the *visual sketchpad*, on which the processing of non-verbal material would depend. The phonological loop is considered to play a key role in vocabulary acquisition, particularly in the early childhood years (Baddeley et al., 1998). The phonological loop would consist of two components: a phonological store served by an articulatory rehearsal component (articulatory loop) process. Within the phonological store, mnestic traces (e.g. of letters heard or images seen) disappear within a maximum of two seconds. If, however, the reiteration process intervenes, it is possible to retain the trace for longer because the information thus replayed can re-enter the phonological store. The process of articulatory repetition would also allow the written, verbal material to be translated into a phonological code that is then sent to the phonological store. Auditory material, on the other hand, would have direct access to the phonological store (Ladavas and Berti, 2014).

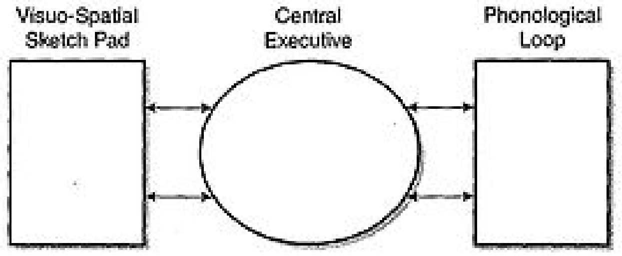


Figure 20. The earlier version of the Baddeley's model of Working Memory.
(Source: Baddely and Hitch, 1974).

The episodic buffer was the last component to be added to Baddeley's working memory model (figure 21). The existence of this fourth component is because, although the tripartite model has been successful in giving an integrated account not only of data from normal adults but also neuropsychological, developmental and neuroimaging data, there are, however, several phenomena that are not readily captured by it (Baddeley, 2000). In fact, the tripartite model predicts that verbal information of the phonological cycle is stored according to a purely phonological code, not explaining the advantage in WM performance when the material presented for the recall is characterised by meaning relations between the stimulus words or the fact that the recall of unrelated words may be influenced by a range of long-term memory aspects such as verbal frequency and imaginability (Roodenrys et al., 2002; Ladavas and Berti, 2014; Baddeley et al., 2021). In addition, studies on amnestic patients show that although they have a very poor performance in recalling prose words, they can produce a near-perfect immediate recall of the exact prose words (Baddeley and Wilson, 2002).

The episodic buffer is therefore defined like a limited capacity passive system. It is a temporary *multimodal store* that combines information from the phonological loop and visuospatial sketchpad subsystems of WM with information about time and order of the presentation to form and maintain an integrated, detailed representation of a given stimulus or event, that can then be deposited into long-term memory as necessary. It is 'episodic' in the sense that it holds integrated episodes or scenes and a 'buffer' to provide a limited capacity interface between systems using different representational codes. In addressing certain shortcomings of the original working memory model—particularly the failure to explain the process of chunking and the dilemma of linking the two distinct representational formats of the loop and sketchpad—the episodic buffer provides a means to allow multiple sources of information to be considered simultaneously, thus creating a model of the environment that may be manipulated to solve problems and plan future behaviour (Baddeley et al., 2010; Baddeley, 2000). So, it can be conceived as a multidimensional storage system that integrates information from different sources, such as long-term memory (LTM), slave systems, and perception (Nobre et al., 2013).

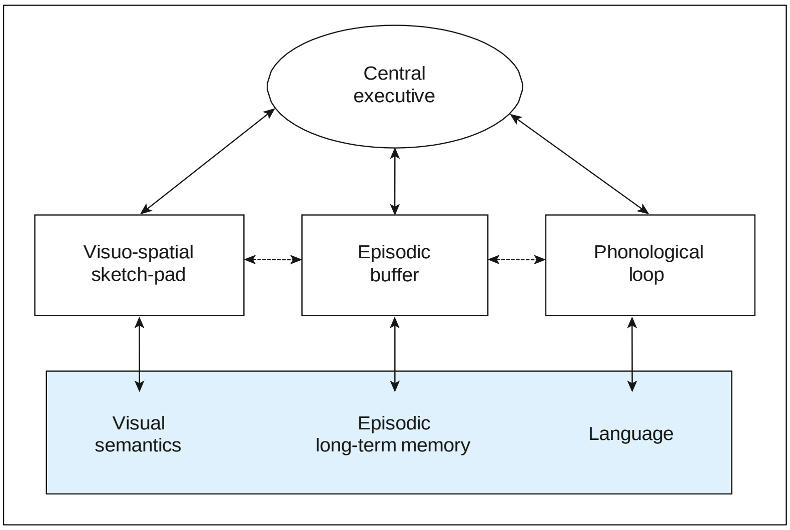


Figure 21: A later development of Baddeley's WM multicomponent model. It includes links to long-term memory and a fourth component, the episodic buffer that is accessible to conscious awareness. (Source: Baddeley, 2010).

As reflected by the fact that it has been labelled as 'the hub of cognition' (Haberland, 1997) and proclaimed as "perhaps the most significant achievement of human mental evolution" (Goldman-Rakic, 1992), WM is a central construct in cognitive psychology and, more recently, in cognitive neuroscience (Miyake and Shah, 1999). Since the publication of the concept of WM around 40 years ago, it has proved to be surprisingly durable (Baddeley, 2000; Baddeley, 2003; Baddeley, 2017; Logie et al., 2020a); (Cowan, 2022). In fact it continues to be actively used within many areas of cognitive science, including mainstream cognitive psychology (Hitch and Logie, 1996; Cowan, 2023) neuropsychology (Becker, 1994; Cubelli et al., 2023), neuroimaging (Smith and Jonides, 1997; Carpenter et al., 2000; Owen et al., 2005) developmental psychology (Hitch and Halliday, 1983; Gathercole et al., 2004; Cowan and Alloway, 2009); computational modelling (Lemaire and Portrat, 2018; Burgess and Hitch, 2005) and hearing diseases in both children (Heinrichs-Graham et al., 2021; Arfé et al., 2014) and elderly (Zarenoe et al., 2017; Na et al., 2017).

To realise the importance of WM, just consider that scientific research shows that it provides a mental workspace used in many essential activities in learning during life, including literacy (Siegel and Ryan, 1989; Gathercole et al., 2006); reading (Cain et al., 2004) and numeracy (Geary et al., 2004). In particular, results support the notion that growth in WM is a significant predictor of children's problem solving beyond the contribution of reading, calculation skills, and individual differences in phonological processing, inhibition, and processing speed (Swanson et al., 2008). Furthermore, Alloway and Alloway (2010) found that working memory at the start of formal education is a more powerful predictor of subsequent academic success than intelligence quotient (IQ). This result has important implications for education, in particular with respect to educational and rehabilitating intervention. Moreover, impaired WM is observed in many neuropsychiatric conditions, such as traumatic brain injury, stroke, mental retardation, and schizophrenia (Ericsson et al., 1980; Klingberg, 2010). Furthermore, deficits in WM are a common feature of a wide range of developmental disorders and specific learning difficulties, including ADHD, dyslexia, specific language impairment (Archibald and Gathercole, 2007), reading and mathematical difficulties (Holmes and Gathercole, 2014; Geary et al., 2007; Swanson and Kim, 2007).

Considering the connection between WM deficits and learning and language impairments, immediately appears as interesting the investigation of the assessment of WM in patients typically affected by such difficulties, that is, hearing impaired persons.

I. 5. 1. Auditory Experience and Working Memory

As introduced, the brain is a highly interconnected information-processing system that develops based on complex interactions between neural activity and sensory stimulation from the environment, including auditory stimulation. This complexity results, as mentioned earlier, in the evidence that deprivation in early auditory experiences and activities may influence the development of more basic elementary neurobiological and cognitive functions extending well beyond spoken language skills (Kronenberger et al., 2014). In fact, because the brain is an integrated functional system, sensory processing (and, by extension, the effects of sensory deprivation) are not entirely independent from the rest of neurocognition and thus may have secondary effects on the brain and cognition as a whole (Conway et al., 2009). Moreover, the temporal patterns provided by the early auditory experience to the developing brain may be necessary for developing sequential processing abilities such as sustained attention and memory for serially presented items. Sound, precisely because of its characteristics as both a temporal and sequential signal hearing (Hirsh, 1967) by providing vital exposure to events ordered serially, encourages the development of sequential processing and behaviour. The sound thus provides a 'scaffold' - a support structure - that organisms use to learn to interpret and process sequential information (Conway et al., 2009), skills as seen, underpinned by working memory. So, because early auditory experience and verbal skills are part of a more extensive functionally integrated system of neurocognitive processes (Geers and Moog, 1987; Luria, 1973); as evidenced by the behavioural research of Pisoni and his group (Pisoni et al., 2008; Pisoni et al., 2012) language delays and/or compromised early auditory experiences may affect other neurocognitive abilities. For example, auditoryï verbal experiences and skills have been implicated in the development of executive functioning ð processes used for directing and controlling thought and behaviour, see (Baggetta and Alexander, 2016) for in-depth conceptualizationð because they provide tools and experiences (such as language and phonological representations of speechï sound sequences) that are used in developing self-regulation, verbal mediation and working memory (Conway et al., 2009).

In the light of the considerations so far, it is clear how language delays and/or compromised auditory experiences may affect neurocognitive abilities. Therefore, the establishment of early neurocognitive function (such as WM) is inextricably impacted by prelingual deafness as well as compromised auditory experience (Beer et al., 2011). Children who experience an early period of auditory deprivation are especially vulnerable to disturbances and delays in verbal short-term and WM processes, including rehearsal and retrieval of verbal information (Pisoni et al., 2012) and deficit in phonological storage capacities (Lyxell et al., 2008). On the other hand, a growing body of work suggests that individuals with smaller WM capacity may be more susceptible to an altered acoustic signal (Arehart et al., 2015). Finally, considering hearing loss in later life (presbycusis described above), there is a broad consensus that verbal WM decreases with age (Moberly et al., 2017; Angelopoulou et al., 2021) and that it is significantly involved in speech recognition in both hearing and hearing impaired adults (Bopp and Verhaeghen, 2005; Nittrouer et al., 2016; Rönnberg et al., 2013).

I. *5. 1. 2. Cochlear Implant and Working Memory*

People with CIs show wide variability in language development performances. In fact, despite deaf adults and children are generally able to achieve excellent speech and language outcomes with CIs, demonstrating the efficacy of cochlear implantation as a medical intervention for the treatment of profound hearing loss, there remain several challenges in the field that deal with the effectiveness of CIs in individual patients (for a recent and interesting review see Tamati et al., 2022).

It is becoming clearly evident in the international field of CIs research that the individual differences and variability in speech and language outcomes following implantation are not merely an "ear issue" that deals with the sensory coding and processing operations reflecting the upstream contributions from the peripheral auditory pathways (Pisoni et al., 2017): the sources of that variability will not be understood until interactions between spoken language and cognitive functions can be taken into account (Nittrouer et al., 2013). The enormous variability in outcomes reflects numerous complex neural and cognitive processes that depend heavily on the functional connectivity of multiple brain areas working together as an integrated system (Luria, 1973).

As Oliver Sacks (2011, p. 133) pointed out

"*The brain is more than an assemblage of autonomous modules, each crucial for a specific mental function. Every one of these functionally specialized areas must interact with dozens or hundreds of others, their total integration creating something like a vastly complicated orchestra with thousands of instruments, an orchestra that conducts itself, with an ever-changing score and repertoire*".

And, in the wave of what the neuroscientist expressed, it is possible that the purpose of auditory neuroscience research applied to cochlear implants lies precisely in seeking out the criticalities of specific neural instruments that, for CI users, can perennially influence their orchestral performance and thus their possible repertoire, both in growth and throughout life.

Thus, the challenges of explaining and predicting individual differences and accounting for poor outcomes after implantation are not just problems for the otology and audiology medical fields. Instead, addressing these challenges requires synergy between several disciplines such as cognitive psychology, cognitive science, developmental sciences, linguistics and, as this thesis proposes, neuroscience. To make further progress, the field of CIs would benefit greatly from contributions from multiple disciplines to generate new knowledge on speech and language outcomes in adults and children with CIs and the information-processing mechanisms by which underlying factors contribute to outcome variability. Such knowledge will provide a scientific basis for developing novel clinical assessments and rehabilitation protocols to optimise outcomes in adult and paediatric CI patients. Specifically in children, previous findings on delay associated with the afore-mentioned Executive Function EF,—namely higher-order cognitive functions that make it possible to formulate objectives and plans, remember these plans over time, choose and start actions that allow us to reach those goals, monitor the behaviour and adjust it to arrive at those goals (Aron, 2008) —are consistent with the hypothesis that a period of profound deafness during critical early periods of brain development, combined with degraded underspecified auditory experience following implantation, may impact broad, domain areas of neurocognitive development (Figueras et al., 2008; Pisoni et al., 2012; Sharma and Campbell, 2011). Of the three broad EF areas (WM, fluency-speed, inhibition-concentration) identified by Kronenberger and colleagues (Kronenberger et al., 2013), as at-risk in long-term CI users, WM mainly was investigated, showing associations with language in children with CIs. In fact, verbal-phonological WM capacity is significantly associated with speech perception skills (Cleary et al., 2000; Nittrouer et al., 2013), grammar (Willstedt-Svensson et al., 2004), vocabulary and reading (Lyxell et al., 2008; Fagan et al., 2007; Köse et al., 2022; Geers et al., 2013), novel word learning (Willstedt-Svensson et al., 2004), and conversational communication (Ibertsson et al., 2009) in youth with CI.

Moreover, experimental studies show that pre-schoolers with CIs manifest significantly poorer performance on inhibition, concentration and WM than peers with Normal Hearing (NH) and national norms (Beer et al., 2014). Therefore, WM's storage appears to be impaired in CI children attending school (Nittrouer et al., 2013). Thanks to Nittrouer and colleagues' longitudinal studies (e.g. Nittrouer et al., 2017) it has been indeed possible to observe that verbal WM deficits in CI children arises due to signal degradation, which limits their abilities to acquire phonological awareness, resulting in a hindering of their abilities to store items using a phonological code.

Reflecting on the medical-logopaedic outcomes considered for the effectiveness of cochlear implantation, even those CI children who can accurately identify speech signals in isolation, may not have phonological WM mechanisms or processing strategies developed similar to age matched normal hearing (NH) children (Cleary et al., 2001). A recent pilot study reveals that CI users show more dual-task interference (decline in speed during the WM condition compared to the standard condition) than NH peers, indicating that their lexical access speed was more dependent on the engagement of WM resources (Kronenberger et al., 2018). In fact, children with CIs have deficits in storing and processing verbal information in WM. These deficits extend to receptive vocabulary and verbal reasoning and remain even after controlling for the higher maternal education level of the NH group (Davidson et al., 2019). Despite the evidences mentioned above, according also to Pisoni and colleagues (Pisoni and Cleary, 2003; Pisoni and Geers, 2000), one of the most important problems in the field of paediatric CI is understanding the enormous individual differences in performance among children on a wide variety of outcome measures that assess speech perception, language comprehension, speech intelligibility and reading. Investigating the causes of this enormous variability in the outcomes of cochlear implant wearers is the challenge facing neuroscience applied to auditory pathology. Two works I have conducted and presented in the experimental research part, are precisely within this multidisciplinary challenge and sought to answer two specific scientific questions (A and B).

I. 5. 2. Brain Imaging and Working Memory

The fast development of neuroimaging techniques over the last few decades has made it possible to obtain various information about the processes occurring in the human brain, both in static situations for diagnostic purposes and in active situations involving the performance of cognitive tasks. In fact, neuroimaging techniques, are used to image the structure and function of the nervous system for medicine, psychology, and neuroscience research (Erol and Hunyadi, 2022).

I. *5. 2. 1. But what is it meant by Brain Imaging?*

A brain imaging (neuroimaging) method could be defined as any experimental technique that allows human (or animal) brain structure or function to be studied, preferably in vivo, in the current context (Brammer, 2009). Conceptually, neuroimaging techniques can be classified into two approaches: structural and functional. *Structural* imaging refers to approaches that are specialised for the visualisation and analysis of anatomical properties of the brain, and they are particularly useful for detecting brain damage and abnormalities. In contrast, *functional* imaging is used to identify brain areas and underlying processes associated with performing a particular cognitive or behavioural task. Depending on the type of signal being analysed, inferences between the location of brain activity and brain function can then be provided (Hirsch et al., 2015). While numerous neuroimaging techniques exist, it is essential to realise that each possesses own strengths and weaknesses. In this context, the concept of resolution needs to be highlighted. Above all, because it was precisely the different temporal/spatial characteristics—as well as further specific details that will be discussed later— that led me to choose specific methods of analysis for the experiments I carried out in my research.

Spatial resolution refers to the ability of an imaging technique to distinguish between two points in space (or structures within the brain) that are nearby. The higher the spatial resolution, the finer the detail that can be provided. In contrast, *temporal resolution* refers to the ability to distinguish between two events occurring over a given time period. The higher the temporal resolution of an imaging technique, the better it can discriminate between two events occurring in close sequence (Hirsch et al., 2015).

Functional neuroimaging techniques allow the examination of human brain function and have revolutionised the way we study neuroscience in humans. It is now possible to probe how the brain works under highly controlled experimental conditions (Ward, 2013). Such a method should ideally produce images in accurate timing and spatial localisation of cerebral function, structure, or changes in these properties of the brain. The most common widely used non-invasive functional methods are electroencephalography (EEG), which, as we will see in detail in the dedicated section of this paper, records electrical activity, while magnetoencephalography (MEG) records the electromagnetic field generated by this electrical activity. Positron Emission Tomography (PET) exploits the information provided by the increased glucose consumption of neurons during their functioning. Finally, Functional Magnetic Resonance Imaging, (fMRI) records the cerebral haemodynamic response resulting from the cerebral haemodynamic response resulting from the increased activity of a particular neuronal population (Babiloni et al., 2007a). It is precisely the characteristics of temporal variability that are particularly attractive in the choice of EEG or MEG compared to fMRI or PET techniques, as the latest have a temporal resolution on the scale of several seconds or minutes, totally insufficient to follow the evolution of brain activity, which instead changes on time scales of the order of tens of milliseconds. In fact, magnetoencephalography (MEG) offers the advantage of a very high temporal resolution (down to tenths of a millisecond) and good spatial resolution (2-3 cm). Electroencephalography (EEG) has a similar temporal resolution, on the order of milliseconds, but has a shallow, spatial resolution (6-9 cm), which is not suitable for studying the mass activity of relatively small cortical areas (Nunez and Cutillo, 1995; Babiloni et al., 2004; Babiloni et al., 2007b). (However, as discussed below, the low spatial resolution of the EEG is not fully confirmed).

Neuroscientific research that has delved into the brain mechanisms of WM has occurred over time in the international scientific landscape. In fact, in the 1980s, studies show that in primates, WM was associated with elevated and sustained neural firing over a delay when information is kept in mind (Funahashi et al., 1989). Neuroimaging studies in humans (using, for example fMRI or electroencephalography (EEG) have mapped WM-related activity to both sensory association cortices and prefrontal cortex, and some of these regions show specificity to the sensory modality of the stimuli (Linden, 2007; Curtis et al., 2003). Other brain regions, including parts of the intraparietal cortex and dorsolateral prefrontal cortex, are activated across several modalities and thus reflect a multimodal type of activity (Klingberg et al., 1996). Moreover, neural network models have suggested that stronger frontoparietal synaptic connectivity is one potential mechanism behind WM development capacity during childhood (Edin et al., 2009). To examine what brain regions are commonly and differently active during various WM tasks (e.g. n-back task[7], the Sternberg task[8], delayed matching to sample-DMTS[9], delayed simple matching-DMS[10]), Rottschy and colleagues (2012) performed a coordinate-based meta-analysis over 189 fMRI experiments on healthy subjects. The main effect yielded a widespread bilateral front-parietal network. Further, results revealed that several regions were sensitive to specific task components, e.g. language Broca's region was selectively active during verbal tasks. At the same time, ventral and dorsal premotor cortex were preferentially involved in memory for object identity and location, respectively. Moreover, the lateral PFC showed a rostral and caudal part division based on differential involvement in task-set and load effects.

Dwelling briefly on EEG technology, there are numerous reports on the modulation of theta band activity in human performing WM tasks (Klimesch, 1999; Gevins et al., 1997; Doppelmayr et al., 1998; Kahana et al., 1999; Tesche and Karhu, 2000; Raghavachari et al., 2001).

Gevins and colleagues (1997) using the 'n-back paradigm' have demonstrated that WM engagement is sufficient for inducing frontal theta activity, but it is unclear which components of the task (encoding, retention or recall) induce the activity. Furthermore, Raghavachari (Raghavachari et al., 2001) applying subdural electrodes in epileptic patients, showed that theta activity persists during WM retention. However, it remains unknown to what extent the intracranially recorded theta activity contributes to the frontal theta activity and what extent these results generalise to healthy subjects. An interesting study by Petersen and colleagues, (2015) investigated whether hearing loss in older participants affects neural mechanisms of WM during an auditory Sternberg task implemented by varying degrees of memory load

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[7] N-Back task 'is one of the most popular experimental paradigms for functional neuroimaging studies of working memory in which subjects are asked to monitor the identity or location of a series of verbal or nonverbal stimuli and to indicate when the currently presented stimulus is the same as the one presented n trials previously (Owen et al., 2005). For more details, please refer to studies I and II of the empirical part of this thesis as the cognitive task was used in the experimental protocol.

[8] The 'classic Sternberg working memory task' involves the presentation of a list of items to memorize, followed by a memory maintenance period during which the subject must maintain the list of items in memory. The maintenance period is terminated by the onset of a 'probe' letter, to which the subject must respond whether the item was in their memorized list of items or not (see Sternberg, 1969).

[9] In a 'Delayed matching-to-sample (DMTS)' procedure, a subject is presented with a sample stimulus. Completion of an observing response to the sample stimulus terminates sample presentation and initiates a delay (usually called the retention interval) between sample offset and the onset of comparison stimuli. In the case of identity matching, a response to the comparison stimulus that matches some physical property (e.g., hue) of the previously presented sample stimulus results in the delivery of reinforcement, and responses to a comparison stimulus that does not match the sample results in a timeout (see Kangas et al., 2011).

[10] Delayed Matching to Sample (DMS)' assesses both simultaneous visual matching ability and short-term visual recognition memory, for non-verbalizable patterns. The participant is shown a complex visual pattern, that is both abstract and non-verbal (the sample), followed by four similar patterns, after a brief delay. The participant must select the pattern which exactly matches the sample. In some trials the sample and the choice patterns are shown simultaneously, in others there is a delay (of 0, 4 or 12 seconds) before the four choices appear (see Amit et al., 2013).

and background noise. Their findings show that participants suffering from a higher degree of HL exhibited a breakdown in alpha activity with increasing task difficulty, which was not observed for the participants with mild or no HL. These results suggest that adaptive neural mechanisms for coping with adverse listening conditions break down for higher degrees of HL, even when adequate hearing aid amplification is provided. Studying MEG responses in healthy subjects performing the Stenberg task to investigate the dependence of frontal activity on memory load (Jensen and Tesche, 2002), it has been possible to show that theta activity increases parametrically with memory load and the activity in this frequency band is sustained during the detention period. These results suggest that theta oscillations generated in frontal brain regions play an active role in memory maintenance. Klimesh's investigation (1999) correlates electroencephalographic components to mental efforts, finding that an essential parameter of the subject's engagement in the task is the "mental workload". This mental workload, also applied in WM tasks (Berka et al., 2007), could be indexed as the ratio between the EEG power spectral density (PSD) in the theta frequency band (which usually increases with the increase of task demand) over the PFC and frontal cortical areas and the EEG PDS in the alpha band (which usually decreases with the increase of task demand) over the parietal cortical areas (Cartocci et al., 2015).

I. 6. Attention

Compared to working memory, the concept of attention can be trickier to frame (Cowan, 1998). The experimental psychologist William James already referred to attention by stating, with great intuition, as follows:

"*Everyone knows what attention is. It is the taking possession by the mind, in clear and vivid form, of one out of what seem several simultaneously possible objects or trains of thought. Focalization, concentration, of consciousness are of its essence. It implies withdrawal from some things in order to deal effectively with others, and is a condition which has a real opposite in the confused, dazed, scatter brained state which in French is called distraction, and Zerstreutheit in German*" (James, 1890, p. 404).

Furthermore, memory and attention figure in almost every cognitive act and, because WM was associated with components of attention control such as response suppression (Kane et al., 2016), goal maintenance (Kane et al., 2001), inhibitory control and protection (Long et al., 2002), to understand one, one must also understand the other (Moscovitch and Umiltà, 1990), although attention was not the main focus of neurophysiological investigations in the present work.

Searching for an operational definition, attention is the function through which it is possible to regulate the activity of cognitive processes by filtering and organising information from the environment to emit appropriate environmental responses. Since the processing system has a limited capacity, the function of attention is to regulate the activity of mental processes by filtering and organising information from the environment. The process of information processing is indeed extremely flexible, i.e., it chooses from time to time what information to process and how to process it. This possibility of selecting information material occurs precisely based on attentional mechanisms (Ladavas and Berti, 2014). Experimental psychology has always supported the identification of several components in the function generically referred to as attention among them alertness, selectivity, and processing capacity (Bagnara, 1984; Umiltà, 1994; Posner and Boies, 1971). Posner and Petersen (1990; 2012) with the aim to integrate behavioural, systems, cellular, and molecular approaches to common problems in attention research, proposed a model of the discrete anatomical basis of the attention system: divided into three networks (figure 22), each representing a different set of attentional processes.

1) The *alerting network* that, through the noradrenergic system in the brain (locus coeruleus), prepares a state of alertness by increasing the speed of elaboration of selected information. This function seems to be lateralised in the right hemisphere.

2) The *orienting network* focuses on prioritising sensory input by selecting a modality or location. Consensus in the imaging literature now indicates that frontal as well as posterior areas are involved in orienting (Corbetta et al., 1998; Thompson et al., 2005). At the cerebral level, the posterior parietal cortex, the pulvinar and the superior colliculus are part of it, each with a specific function respectively of uncoupling, coupling and shifting attention.

3) Finally, the *executive network* (the anterior attentional system) presides over target detection and awareness. Although it is possible to monitor for targets in many processing streams without too much difficulty, the moment of target detection produces interference across the system, slowing the detection of another target (Duncan, 1980). This set of process is related to the limited capacity of the attention system and to awareness itself and has often been called *focal attention*. One might think of focal attention as the entry to the conscious state, which may involve widespread connections from the midline cortex and the anterior cingulate cortex (ACC) to produce the global workspace frequently associated with consciousness (Dehaene and Changeux, 2011). Target detection and awareness connected to the medial frontal cortex and the adjacent ACC. This brain region has been highly studied by imaging experiments partly because of its frequent activation (Petersen and Posner, 2012).

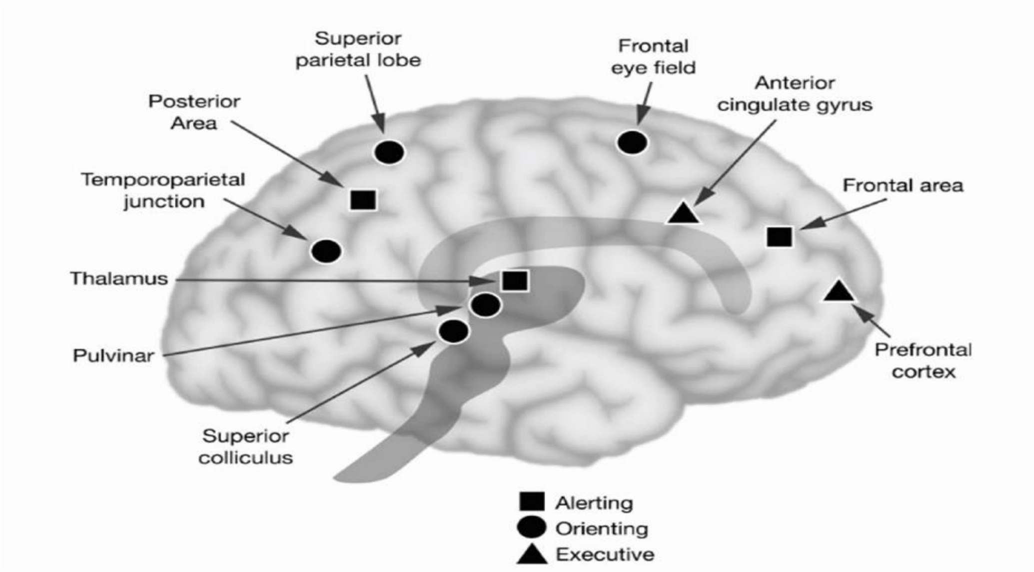

Figure 22. The neuroanatomy of attentional networks. The figure illustrates the cortical areas involved in the three attention networks. The alerting network (squares) includes thalamic and cortical sites related to the norepinephrine system. The orienting network (circles) is centred on parietal sites, and the executive network (triangles) includes the anterior cingulate and frontal areas. (Source: Posner and Rothbart, 2007).

It emerges from this brief mention of the construct of attention how it is a cognitive function linked to the functioning of different cortical areas and closely connected to the sensorial cues that stimulate the brain from the outside world. This interaction between attention and the sensory systems emerges clearly from the words of Treisman, a pioneer in the study of attention, who defined it as the selective aspect of perception and response (Treisman, 1969). Therefore, it is intuitable how its efficiency is strongly linked to the sensory systems that transfer the sensorial message to the brain allowing for perceptual processing. Clearly, if sensory deficits are not present, attention, especially selective attention that filters distractors and selects the target information to be processed and stored in working memory (Cotton and Ricker, 2022), can act and support cognitive functioning. However, what happens if sensory systems, such as hearing, are impaired? Clearly, in the light of the interconnections described above, hearing impairment treated with cochlear implants can lead to cognitive problems in attention and/or working memory, both when it occurs at the beginning of the wave of life and on its crest and descent (presbycusis).

I. 6. 1. Attention in Hearing

Some of the earliest work examining human selective attention considered auditory communication signals, focusing on the issue of how we choose what to listen to a mixture of competing speech sounds (Shinn-Cunningham et al., 2015). Auditory selective attention refers, in fact, to the mental ability to resist distractor stimuli and select relevant information from the surrounding acoustic events (Giard et al., 2000), as illustrated in the 'cocktail party effect'. The first experimental studies on auditory selective attention are due to Cherry in healthy subjects (Cherry, 1953) and children with learning disabilities

(Cherry and Kruger, 1983). Furthermore, hearing loss occurring with advancing age, as is sadly well known, is associated with a slowdown in cognitive processing and a decline in attentional resources (Wingfield et al., 2005; Pliatsikas et al., 2019). As will be shown, it was the attentional mechanisms in the elderly and children with cochlear implants that I investigated using psychometric instruments to answer two scientific questions (F and G) in the part III of the thesis.

I. 7. Emotional Waves

Despite Fehr and Russell (1984) highlighted the difficulty in producing an explicit definition of emotion, observing that *"everyone knows what an emotion is until asked to give a definition. Then, it seems, no one knows"*, cognitive and affective neuroscience can significantly contribute to this effort with an understanding of emotion as a scientific concept, in particular by offering functional architectures of emotional processes in the form of explicit models (Sander, 2013).

In the categorial basic emotion model, the adjective "basic" is used to express three postulates (Sander and Koenig, 2002); (Ekman, 1992a) for reviews see (Brosch et al., 2010) and (Ortony and Turner, 1990). First, to convey the notion that there are several separate emotions which differ one from another in meaningful ways. Second, the term refers to the notion that non-basic emotions are made up of blends of basic emotions (e.g. Tomkins, 1963). Third, the term is used to indicate that evolution played an essential role in shaping both the unique and the standard features which these emotions display and their current function. Finally, basic emotions can be defined as "affect programmes" that are triggered by appropriate eliciting events to produce emotion-specific response patterns such as prototypical facial expressions and physiological reactions (Ekman, 1992a) driven by specific neural response systems (Panksepp, 1998).

Although the exact set of emotions considered basic and their definitions vary among theorists (Armory and Vuilleumier, 2013; Keltner et al., 2019), the paradigmatically basic emotions are anger, disgust, fear, joy, sadness, surprise and neutral that could be recognised cross-culturally (Ekman and Friesen, 1971; Fridlund et al., 1987), (for a recent "cultural evolutionary" perspective see (Lindquist et al., 2022). On the other hand, according to the dimensional perspective, there are a few fundamental dimensions that organise emotional responses (Mauss and Robinson, 2009). The most commonly assumed dimensions are *arousal* that concerns the subjective perception of perceiving energy intensity (high-low), *valence* (sometimes referred to as activation), that corresponds to the level of perceived positivity/negativity (Russell, 1980) and *approach-avoidance,* characterised by tendencies to approach stimuli (e.g., as would likely be facilitated by excitement), whereas avoidance motivation is characterised by tendencies to avoid stimuli (e.g., as would likely be facilitated by anxiety) (Davidson, 1999; Lang et al., 1997; Schneirla, 1959; Reznik and Allen, 2018).

Moreover, emotions play a critical role in the evolution of consciousness and the operations of all mental processes (Izard, 2009). So they occur in the context of ongoing social relationships interacting with internal cognitive, motivational and physiological components that characterise each individual: emotion knowledge is, therefore, a relevant component of social competence. (Giménez-Dasí et al., 2013). Numerous studies have shown a close relationship between emotion comprehension and social competence from very young ages through adolescence (Mostow et al., 2002; Denham et al., 2003). Specifically, during life, humans use and develop a range of cues to communicate emotions, including vocalisations, facial expressions and posture (Coulson, 2004; Ekman and Friesen, 1980); so emotional maturation is a complex and gradual process that starts in early years and continues until adulthood (Mancini et al., 2016). Converging evidence suggests that the ability to discriminate visual cues like facial expressions emerges early in infancy: by five months infants habituate to faces showing the same expression across differing identities (Bornstein and Arteberry, 2003); by seven months, infants can discriminate a habituated facial expression (happy) from fearful and angry faces (Kestenbaum and Nelson, 1990). Moreover, infants are sensitive to vocal cues from the beginning of life, when their visual system is still relatively immature (Mehler et al., 1978); (Inguscio, 2015).

Affective prosody (changes in pitch, loudness, rate, and rhythm of speech to convey emotion) communicates the speaker's emotions and social intent (Patel et al., 2018). In fact, auditory signals allow for effective communication when the recipient cannot see the sender, for example, across a distance or at night (Sauter et al, 2010). Although vocal expression of emotions can occur overlaid on speech in the form of affective prosody, humans also use a range of nonverbal vocalisations to communicate how they feel, such as screams and laughs (Sauter et al., 2010). Moreover, the perception of other people's emotions from non-verbal cues is a fundamental component of interpersonal communication) (McArthur and Baron, 1983) and communicating emotion is a fundamental feature of human social interaction that transverses all cultures (Bryant and Barrett, 2008). In fact, the ability at recognising emotions is conceptualised as an adaptive skill in both children and adults, as it should help to infer interaction partners', intentions, anticipate their behaviours, and adapt one's own behaviour in order to achieve interpersonal goals (Halberstadt et al., 2001; Hall et al., 2009; Hampson et al., 2006). Moreover, emotions act upon essential cognitive functions like reasoning, memory, problem-solving and decision making (Tyng et al., 2017; Schmeichel and Tang, 2015). Finally, considering a universal art form such as music, it is important to remember that people appreciate music above all for aesthetic reasons: the emotions it generates, the memories it evokes, and its beauty (Brattico and Pearce, 2013).

I. 7. 1. Emotions and Deafness

In light of what has just been reported regarding the role played by emotions in the development of cognitive abilities, and considering that most research focused on auditory speech perception as a benchmark for implant success (von Eiff et al., 2022), despite the extreme variability in language performances (Geers, 2002), it is pertinent to dwell for a moment on emotional competence in deaf children (and adults) with cochlear implants considering recent research increasingly points to the importance of emotional communication skills in CI users.

First of all, findings from several studies have suggested that deaf children have difficulties with emotion identification, and regulation and that these may impact social skills (Ludlow et al., 2010; Calderon et al., 2011). Moreover, some may argue that emotional cues from the very basis of human interaction contain even more valuable information than spoken words (Jiam et al., 2017). As mentioned, perception of emotions from nonverbal vocal expression is vital to understanding emotional messages, shaping listeners' reactions and subsequent speech production. (Banse and Scherer, 1996). Considering the massive everyday use of communication media, the importance of the vocal communication dimension —concerning, for example, the interlocutor's emotional state, using only auditory cues—emerges. In fact, although facial expressions can be strong indicators of the interlocutor's emotional state, the recognition of vocal emotions is an essential component of auditory-only communication (Luo et al., 2007) (e.g., a telephone conversation, online learning, listening to the radio, podcasts, etc.). Reflecting on this, many are the clues that come into play when communicating an emotion, and one of the most important ones, recognised even in more or less ancient times, is the non-verbal one Cicero's De Oratore cf (Scherer, 1993; Skinner, 1935; Wallbott and Scherer, 1986). Moreover, of all the types of non-verbal cues, humans frequently use prosodic vocal cues (e.g., pitch and timing of voice) to elicit emotional information in their interactions (Planalp, 1996). It is therefore natural that when prosodic vocal cues are degraded, the perception and production of vocal emotions are affected (Jiam et al., 2017). Concerning facial emotion recognition, as early as Darwin (1872) argued that the perception of facial expressions was critical to adaptive responses and behavioural adjustment between peers. The communicative value of facial expressions is thus considered key in human communication. (Ambert-Dahan et al., 2017).

Moreover, the evidence that people (young and adult) with deafness strongly rely on dynamic facial cues for verbal communication both before and after cochlear implantation (Lazard and Giraud, 2017; Lee et al., 2007) raises the issue as to whether this detracts from the perception of nonverbal, emotional, dynamic facial cues (Ambert-Dahan et al., 2017). Effectively, findings from several studies suggest that deaf children are more prone to errors in recognising facial expressions of emotion than their hearing controls. In particular, people with prelingual hearing loss make more errors than people with postlingual hearing loss (Bachara et al., 1980; Schiff, 1973), suggesting a sensitive period for emotional competence as well. However, later studies indicated that deaf children could perform as well as hearing children on a simple emotion recognition task when it involves emotion *matching* rather than emotion *recognition* (Hosie et al., 1998; Weisel, 1985).

Furthermore, studies have found that the language level of normal-hearing children (NH) is linked to their capacity to recognise emotions in social contexts (Sidera et al., 2017) so NH children with a language delay could also experience difficulties in identifying the characters' feeling in short stories with pictures of faces depicting emotions (Nelson et al., 2011). Furthermore, scientific evidences suggest that children's difficulty in understanding mental states in general and to recognize emotions in particular facially, *is not* due to problems with neurocognitive structure (as is sometimes attributed to children with autism) but to problems with language acquisition (Dyck and Denver, 2003; Ludlow et al., 2010; Spencer and Marscharl, 2010). This explanation is supported by the fact that deaf children's deficit in social cognition (Theory of the Mind see Gallese et al., 2004 and Baron-Cohen et al., 1985) occurs only in those (typically born to hearing parents) who do not have access to a language from birth (Schick et al., 2007; Dyck and Denver, 2003). In fact, as seen previously, language acquisition is an essential ability that occurs more naturally and successfully in the first years of life (Gervain and Mehler, 2010; Lenneberg, 1967). Hence, the capacity to acquire language diminishes with age and some aspects of language cannot be mastered if there is absence of early (sensorial) linguistic input (Humphries et al., 2014). Clearly, access to spoken language depends on the severity of impairment and the timing and quality of audiological intervention (Moeller and Toblin, 2015; Toblin and Moeller, 2015). Therefore, children with deafness may experience months, often years, of sensory and language deprivation (Lederberg et al., 2013), developing deficient emotional-cognitive patterns that will affect their entire development.

I. 7. 2. Measuring Emotions in Neuroscience

Up to now, an attempt has been made to offer an introductory overview of what emotions are and their extreme importance in developing cognitive and social skills in humans. However, given the importance of their knowledge, the question arises as to how they can be measured and quantified in the field of neuroscience. Clearly, depending on the observation perspective adopted (discrete, therefore single emotions; dimensional global activations such as arousal valence), different instruments are used in research.

- *Self-Report measures* allow people's perceptions to be recorded online. However, individual differences in awareness of and willingness to report on emotional states potentially compromise even online reports of emotional experiences. These differences may be due, for example, to the social desirability of the subject being measured (Paulhus and John, 1998) or to deficits concerning emotional processing such as alexithymia (Lane et al., 1997).

- *Autonomic Nervous System (ANS) measures.* The most commonly assessed indices of ANS activation are based on electrodermal (i.e., sweat gland) or cardiovascular (i.e., blood circulatory system) responses and pupil dilatation (pupillometry, Rafique, 2020). Electrodermal responding is typically quantified in terms of skin conductance level (SCL) or short-duration skin conductance responses (SCRs). The most commonly used cardiovascular measures include heart rate (HR), blood pressure (BP), total peripheral resistance (TPR), cardiac output (CO), pre-ejection period (PEP), and heart rate variability (HRV). Each of these measures varies in terms of whether it primarily reflects sympathetic activity, parasympathetic activity, or both (Mauss and Robinson, 2009); (see Shu et al., 2018 and Lin, 2023 for reviews on emotion recognition based on physiological signals).

- *Startle response*, an extreme response to a highly novel and/or intense emotional stimulus that carries potential major salience for the intact organism (Costa and Ricci Bitti, 1998) can be used as a marker of the valence dimension of emotional states.

- *Neurophysiological measures*: scientific evidences proposed that the physiological correlates of discrete emotions are likely to be found in the brain rather than in peripheral physiological responses (Buck, 1999; Izard, 2007; Panksepp, 2007). Considering the EEG technique, which will be explored extensively in another section of the present work, the 'frontal asymmetry', which contrasts the alpha-band activation power of the left frontal region with that of the right frontal region, is often referred to in the assessment of emotional response. This asymmetry-based measure has been crucial in the emotion literature (Davidson, 1999) as left activation would be linked to the processing of emotions positivity valanced. Subsequent studies have supported an association of frontal symmetry with approach/avoidance rather than with valence towards an emotional stimulus (Sutton and Davidson, 1997). As will be presented in the experimental part of this thesis, an experimental study (V) assessed precisely frontal asymmetry in response to emotionally charged stimuli in clinical samples. Moreover, MEG studies (Gainotti and Gainotti, 2020; Kheirkhah et al., 2021), demonstrated that the right hemisphere is more involved in responding to emotion than the left hemisphere. Neuroimaging in-vivo studies, applicating technologies such as fMRI (Detre and Floyd, 2000), or PET (Volkow et al., 1997), can localise activation in more delineated brain regions than EEG and thus can detect the specificity of individual emotions in the brain (Panksepp, 1998). Moreover, some studies seem to identify key brain regions particularly related to specific emotions. For instance, the amygdala, a subcortical region necessary for processing stimulus value, is connected to fear (Adolphs, 2008) or, more generally to unexpected inputs of motivational meaning, and ambiguous stimuli (Berridge, 1999; Pessoa and Padmala, 2005; Hulsman et al., 2021).

Another area fundamental to the neurophysiological correlates of emotion is the frontal and prefrontal cortex. Studies on sadness show, in fact, activations in the medial prefrontal cortex and anterior cingulate cortex (Murphy et al., 2003). Finally, when considering anger and happiness, their neural basis a still a matter of debate (Leung et al., 2019; Tanzer et al., 2020).

Thus, the evidence just mentioned seems to suggest that in analysing the neurophysiological correlates of emotions for research purposes, it is important to consider, as for those involving cognitive functions, the global activation underlying emotional and/or cognitive processes by observing it over the time of its manifestation, rather than statically, in search of activations of specific areas. Thus, the temporal resolution allowed by brain signal acquisition techniques, such as EEG and MEG, would allow a broader understanding of activation mechanisms and support a dimensional rather than a punctual approach to the study of emotions. And it was the demand to investigate the electrophysiological patterns of possible emotional deficits in children with cochlear implants that prompted one of the studies (C) that will be presented in the experimental part.

-*Behavioral Expression measures.* One area of research claims that it is possible to deduce a person's emotional state from the behaviour triggered by certain vocal, facial and bodily expressions (Darwin, 1872; Ekman, 1992b).

Regarding *vocal* expressions, the most consistent association reported in the literature is between arousal and vocal pitch, whereby higher arousal levels have been linked to vocal samples with a higher pitch. Based on findings of this type, Bachorowski and Owren (1995) suggested that vocal intonation can be used to assess the level of emotional arousal currently experienced by the individual. Concerning *facial* expressions, supporting the theory that facial expressions had evolved to regulate sensory intake (Darwin, 1872; Susskind et al., 1998), but then these "sensory gatekeepers" were further adapted to serve social signals as well, research suggests that the emotional states behind these expressions may also participate in complex socioemotional interactions (Gardhouse and Anderson, 2013). The study of the physical micro-expressions ME — spontaneous, subtle, and rapid (1/25 to 1/3 s) facial movements reacting to an emotional stimulus (Ekman, 2009; Ekman and Friesen, 1969)— provides the ability to expose genuine emotions that occur briefly and unintentionally, even when true emotions are deliberately masked see (Ekman, 2003) for a review on ME recognition). Moreover, facial behaviours potentially indicative of emotion can also be assessed with facial EMG, which involves measuring electrical potential from facial muscles via electrodes on the face (Dimberg, 1988);. Finally, *body posture* has not received much attention as a measure of emotions. Some findings suggest that the link between emotion and posture may be specific to more culturally related elements, such as social status (Mauss and Robinson, 2009).

II. DETECTION OF BRAIN *WAVES*

Until now, we have analysed the route that an auditory stimulus takes through the mechanical wave and its translations from the air in the environment around us to the encephalon, which processes its meaning through the development of cortical brain activations and connections in and between different brain areas.

These macroscopic activations that have been mentioned are made possible by the microscopic electrical variations that—as will shortly be discussed in more detail—neurons make through the variation of their action potential. And are precisely these variations that, by projecting the signal onto the cortical scalp, allow the recording of the electrical signal, the key to access behind the scenes of the representation of ourselves. So, yes, every action, thought, or movement we produce or process internally generates electricity. Paraphrasing Billy Elliot's response in the exciting film[11], when the panel judges his performance asked what he felt when he danced, *'I feel electricity'*: that is it, we are made of electricity, and therefore, of waves.

Moreover, it is through that electricity that, thanks to the technique of electroencephalography, we can discover the mechanisms and interactions underlying cognitive functions, emotions, thoughts.

II. 1. Why Electroencephalography

Why apply EEG and no other functional analysis techniques such as MEG (Cohen, 1968) or haemodynamic and metabolic correlates of resting-state neuronal activity such as fMRI or PET?

Certainly, as mentioned above, because of their high temporal resolution (milliseconds), EEG and MEG are valuable techniques for the study of brain dynamics and functional cortical connectivity (Babiloni et al., 2004). Indeed, the high temporal resolution offered by both techniques allows to understanding human cognition by analysing brain activations during specific behaviours, such as active tasks of working memory, emotional recognition, speech decoding, and passive tasks such as listening to auditory stimuli or watching images or films, i.e. (Dash et al., 2020); (Costers et al., 2020) see (Beppi et al., 2021) for an up-to-date account of how EEG/MEG oscillations have contributed to the understanding of cognition. Furthermore, in order to answer the experimental questions that arose during my research, the temporal resolution made it possible to conduct experiments that aim to capture fully the rich temporal dynamics that underlie cognitive processes rather than locating with high spatial precision the brain regions engaged in these processes. Moreover, since electrical activity produces, as magnificently summarised by Maxwell's equations (Huray, 2011), not only electric fields but also tiny magnetic fields, it would been also possible to use MEG, which measures extracranial magnetic fields originating from neuronal activity with high temporal resolution and without higher spatial resolution than the EEG as, for some, erroneously

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[11] https://www.youtube.com/watch?v=riH9wfVNlhw

reported (Srinivasan et al., 2007), but this point is still debated (Singh, 2014). A further advantage of MEG over EEG is because the human body is essentially transparent to magnetic fields. So magnetic fields are not distorted by the tissue conductivity of the scalp, skull, cerebrospinal fluid (CSF) and brain. In contrast, electrical fields may be distorted by the skull and CSF (Singh, 2014).

However, two strong pieces of evidence in support of EEG in the context of auditory neuroscience emerge. The *first* concerns the characteristics of the equipment and the fact that a large majority of the subjects involved in the studies covered by this work were minors.

In fact, the EEG allows for an exceptionally eco-friendly, portable and totally non-invasive signal acquisition, using fitted head size adapted commercial caps with adaptable electrode positioning, i.e. (Hairston et al., 2014), completely the opposite of the fixed dimension of the adult MEG helmet, which, if applied to children, resulting in a considerable distance between active neural circuits of the brain: the ‘small head’ problem (Chen et al., 2019).

The MEG, on the other hand, requires a very complex acquisition set-up, as it uses a large number of magnetometers, SQUID (Superconducting Quantum Interference Devices sensors) (Silver and Zimmerman, 1965), several hundred, that, for operation, have to be immersed in liquid helium at the temperature of 4 K (− 269 °C) (Hari and Salmelin, 2012). Furthermore, the exceptionally high sensitivity of the SQUIDs also requires the use of highly sophisticated and expensive shielding systems from external noise sources. Moreover, any head motion relative to the sensors reduces data quality markedly (Boto et al., 2018). All this means that this technique can only be used in a few specialised centres and is not as portable and environmentally friendly as the EEG apparatus (figure 23).

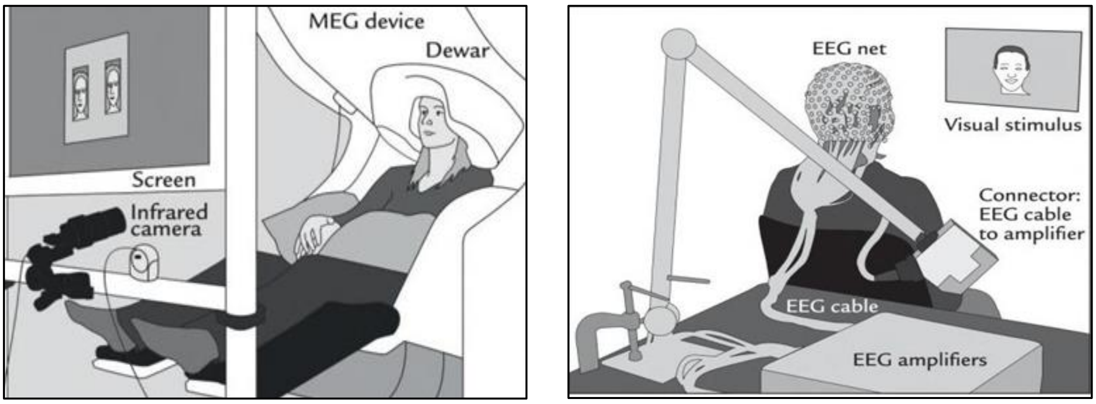

Figure 23. MEG and EEG recording set-up. Schematic MEG layout (right) displays the subject sitting with her head placed in a helmet-shaped in front of her a back-projection screen for visual stimulus presentation. On the right, the EEG set-up shows a subject with attached EEG sensor sitting in front of a computer with a visual stimulation. (Source: modified by Hari and Puce, 2017).

The *second* piece of evidence concerns the applicability. MEG recording from CI users is technically challenging (Debener et al., 2008) and impossible with most CI devices containing, as explained, a strong permanent magnet, which fixes the transmitter coil (Pantev et al., 2006). Furthermore, the presence of electrical and magnetic signals complicates signal acquisition and subsequent processing already in the EEG; it is left to the reader to imagine how complex it can be to identify the source of a magnetic signal in the brain when a magnet and a processor sending electrical signals to the cochlea are present, see (Giraud et al., 2002) for a review on the brain imaging techniques for studies in cochlear implant patients. Finally, electric recording by EEG, as highlighted in the literature, is convenient, widely used, and inexpensive, whereas magnetic recording by MEG is expensive, cumbersome and remains largely confined to specific settings (Nunez, 2012). Therefore, in the light of the above considerations, my experimental investigations mainly used electroencephalography to acquire neurophysiological signals.

II. 2. The Origin and Development of Brain Waves

The discovery of the electrical roots of the brain, like those of the cochlea already described, developed in Italian minds. Were indeed the experiments of the Italian scientists Luigi Galvani (1737-1798) and Alessandro Volta (1755-1832), together with Georg Ohm (1787-1854) and Michael Faraday (1791-1867,) who proved that biological tissue, especially muscle, had many electrical properties. Then, again, an Italian, Carlo Meucci (1811-1868), using Leopoldo Nobili's non-static galvanometer (1784-1835), proved that biological tissue was excitable and directly generated electric current (Mecarelli, 2019a).

The German psychiatrist Hans Berger (1873-1941) was responsible for the first recording of the brain's electrical activity through electrodes placed on the scalp of a 17-year-old boy during surgery on 6 July 1924. The scientist's almost obsessive goal, triggered by a traumatic experience, was to record the "physical basis of the mind", searching for the "energy of mind" (La Vaque, 1999). And it was his mind, probably in a depressive moment without energy, that led him to suicide. Beyond the historical aspect of Berger's discovery, which is not due but, in my opinion, necessary in order to contextualise and realise the humanity behind every technological tool that is often assumed to be processed only by computer,; it is worth remembering how, after five years of further study (Berger, 1929), Berger published the first of 14 articles that established electroencephalography as an essential tool for clinical diagnosis and brain research (Wolpaw and Wolpaw, 2012).

The first EEGs recorded by Berger on photographic paper lasted for 1-3 minutes, and consisted of one EEG channel, with frontal-occipital bipolar derivation, one channel for simultaneous recording of the electrocardiogram and one channel for time making (figure 24).

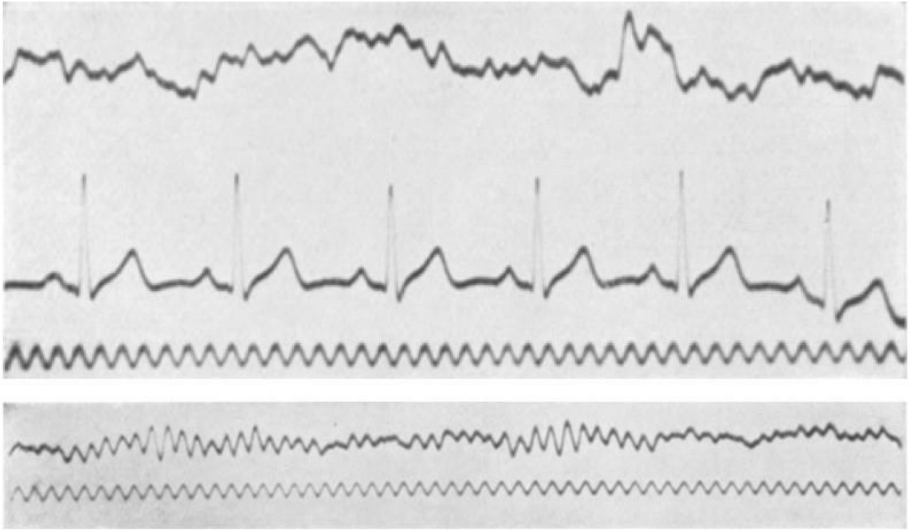

Figure 24. The first EEG was recorded. The first reports on human EEG date back to Hans Berge's first publication (Berger, 1929). Both represent EEG samples recorded by his son Klaus (16). The bottom figure represents a sample of what he would later call an 'Alpha rhythm' (a sinusoidal rhythm of about 10 Hz), and the top figure is what he would later call a 'Beta rhythm' (or a desynchronised EEG with no obvious rhythmicity). The lower trace in both graphs is a sine wave generated at 10 Hz, while the middle trace in the upper figure is the EEG.
(Source: brainclinics.com, 2023).

Already in his first scientific report, Berger listed the characteristics of certain rhythms (which will be described next) by indicating them with the initial letters of the Greek alphabet. H, therefore, identified the Alpha rhythm in the waking state, with eyes closed and reactivity with eyes open, and the Beta rhythm at high frequency. From the 1930s onwards, the science of the EEG evolved rapidly simultaneously with the evolution of the material needed for its recording (Collura, 1995). Finally, as Mecarelli points out, almost 90 years after the publication of the first report on human EEG by Hans Berger, it can be affirmed that electroencephalography maintains its interest and usefulness, both in the diagnostic branch as well as a neurophysiological technique to be used in the experimental fields (Mecarelli, 2019a).

II. 3. Anatomical and Physiological Considerations

Nervous tissue is composed of nerve cells, the *neurons* together with a trophic and supporting tissue called *neuroglia,* essential for providing metabolic support to neurons and for myelinisation of axon fibres (Philips and Rothstein, 2017). Neurons are highly differential and specialised cells, and although they share many of the characteristics of all other cells, they have the unique feature of being able to communicate quickly and precisely with other cells located in distant organs of the body. This property is due to two characteristics: morphological and functional *asymmetry*. They are, in fact, made up of a receptive 'dendrite' on one side and a transmitting 'axon' on the other. In this way, the neuron can transmit unidirectional messages. Secondly, they are excitable both electrically and chemically. The cell membrane of neurons contains a series of specialised proteins, ion channels (sodium Na^+ and potassium K^+) and receptors that facilitate the flow of particular inorganic ions that allow the redistribution of electrical charges by generating currents that change the voltage existing at the ends of the membrane. These potential changes result in the onset of a depolarisation wave that spreads in the form of an'action potential', a special type of electrical signal that moves along its axons: this is the usual way electrical signals travel along the neuron (figure 25). Glial cells, unlike neurons, are less excitable but have the task of supporting and regulating nerve functions (Schwartz et al., 2015).

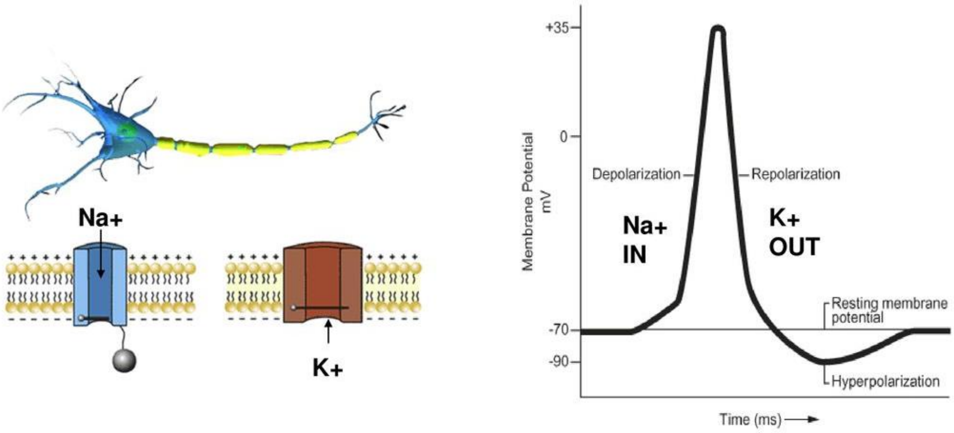

Figure 25. Event of action potential in neurons. The most important ions involved in the action potential are sodium (Na+) and potassium (K+). Each of these ions has its own special voltage-gated channel that opens in response to the depolarisation of the cell membrane. The Na+ channels open quickly, while the K+ channels open later. The movement of the ions across the membrane changes the membrane potential and produces the action potential at the axon hillock. (Source: slideplayer.com, 2023).

So, synthetically, a neuron consists of the cell body (*soma*), which contains the nucleus and much of the metabolic machinery, the dendrites, which are threadlike extensions that receive stimuli from other cells; and the *axon*, a single long fibre that carries the nerve impulse away from the soma to other cells: The soma's morphology characterised the cell type, qualifying it in unipolar, bipolar, multipolar, Purkinje, stellate, pyramidal (Purves et al., 2001) (see figure 26 for a synthetic non-exhaustive representation).

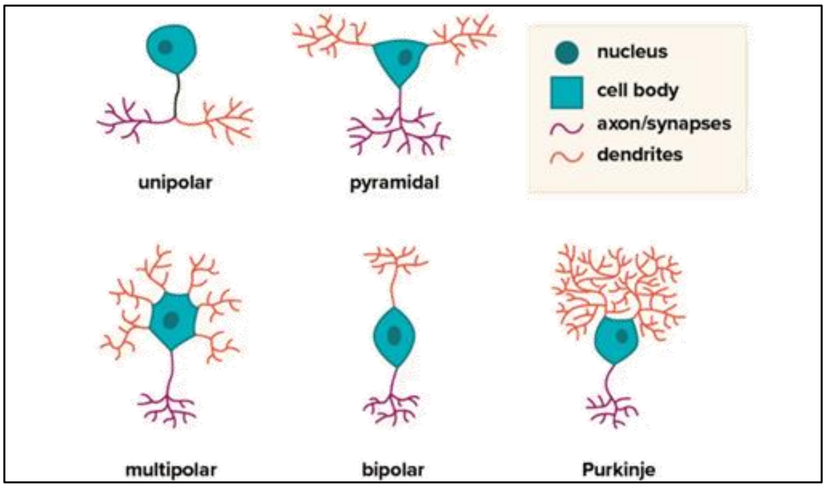

Figure 26. Types of neurons. (Source: healthline.com, 2023).

The two principal groups of cortical neurons, whose consideration is obviously connected to the present work, are the pyramidal and stellate cells (Hämäläinen et al., 1993). The former takes their name from the inverted pyramid shape of their body, are relatively large and most abundant in the cortex, about two-thirds of all cortical neurons are pyramidal. The stellate neurons are relatively small and multipolar. They form about one-third of the total neuronal population of the cortex (figure 27).

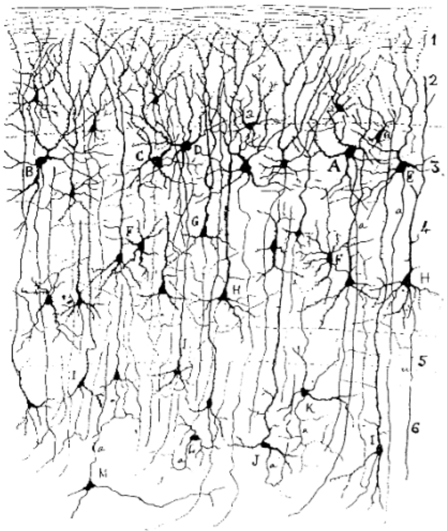

Figure 27. The picture drawn by Ramon y Cajal in 1888 illustrates a prominent pyramidal Neuron (H) in the middle and two stellate cells nearby (F). (Source: Hämäläinen et al., 1993).

Concerning pyramidal neurons, apical dendrites from above reach out parallel to each other, so that they tend to be perpendicular to the cortical surface. The dendrites and the soma typically have thousands of synapses (connections) from other neurons. The intracellular potential is increased by input through the excitatory synapses but decreased by inhibitory input. Most excitatory synapses are on the dendrites; inhibitory synapses often attach to the soma. The neuron fires the action potential when the potential at the axon hillock reaches a certain threshold level (figure 28).

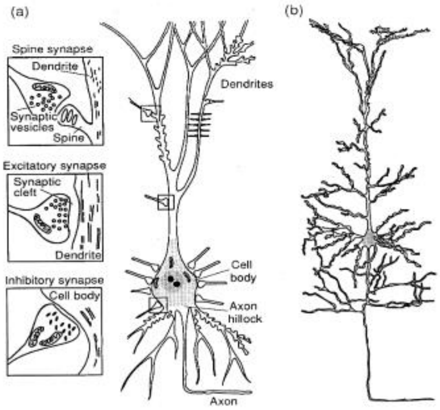

Figure 28. Cortical neuron. (a) Schematic illustration of a pyramidal neuron and three synapses. (b) Pyramidal neuron. (Source: Hämäläinen et al., 1993).

Briefly, this functionality of the neuron is guaranteed by the cell membrane, a selective filter, where specific ion channels are located. The voltage-dependent channels on the presynaptic cell membrane perform a double function: to depolarise the presynaptic membrane, then trigger the action potential and to produce a current such intensity to generate a variation of action potential also in the postsynaptic cell. All the neurons establish interneural connections through the synapses that can be achieved at the cell body level, the dendrites, and the axon (Brienza and Mecarelli, 2019).

According to their transmission mechanism, as anticipated, synapses are divided into chemical and electrical. The former makes communication between cells possible through the secretion of neurotransmitters in the space that separates them (synaptic cleft): chemicals released by presynaptic neurons produce a secondary current flow in postsynaptic neurons by activating specific receptor molecules without there being cytoplasmic continuity between one cell and another. In contrast, in electrical synapses, presynaptic and postsynaptic cells communicate through specialised channels, the gap junctions (Sigelbaum and Kandel, 2019). Nerve transmission through the electric synapses is rapid because it results from the direct passage of the current generated by the voltage-dependent channels of the pre-synaptic cells, with a virtual absent delay. Moreover, this kind of synapse often interconnects entire populations of neurons and, in these cases; the function is to synchronise their responses. When many cells are interconnected by electric synapses, such as in the cerebral cortex, the threshold is exceeded, and the whole group of electrically coupled neurons will tend to discharge synchronously and maximally as the action potential is "all-or-nothing". These synapses are typical of the pyramidal cells, the base of the neural organisation of the cerebral cortex (DeFelipe and Fariñas, 1992), the fulcrum of electroencephalographic studies.

II. 3. 1. Functional Structure and Electrical Activity of the Human Cerebral Cortex

The human cerebral cortex is a 3 to 4-mm thick laminal layer on the brain surface, its development is what most differentiates humans from other species that preceded them phylogenetically. From an anatomical-functional point of view, the organisation of the cerebral cortex is surprising. Each of the six layers in which it is formed contains specialised neurons for receiving and exchanging information between the various regions or for sending out commands to the muscles (Rossi, 2020; Swenson and Gulledge, 2017): as taking up the marine metaphor, a hyper-specialised harbour connected to all the seas of the word. Korbinian Brodmann (1868-1916) first defined the cytoarchitecture of the cortex based on histological and functional differences, which consists of 52 areas (figure 29) that he identified and is still a benchmark for neuroimaging studies (Loukas et al., 2011).

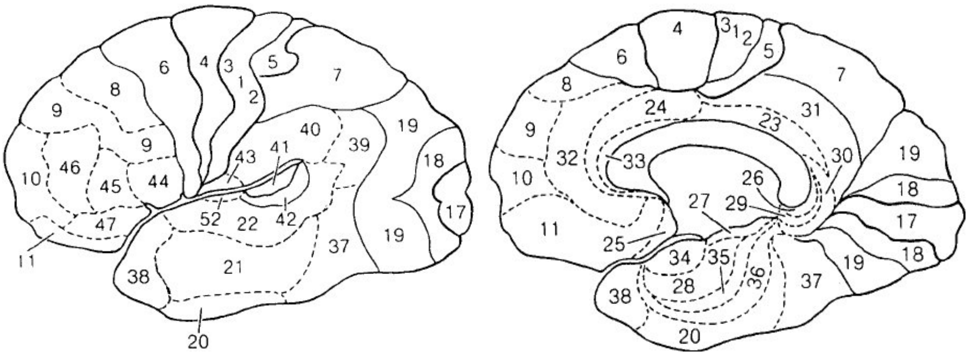

Figure 29. Representation of the Brodmann cortical areas of both hemispheres.
(Source: Cabeza and Nyberg, 1997).

For more on the concepts behind Brodman's cytoarchitectonic maps and reflections on their impact on current neuroimaging approaches, see (Zilles, 2018). Beyond the specific details of the areas —that are not the subject of this part of the work but that have clearly been a reference for the empirical studies that are the subject of the next section of this paper—it is legitimate to ask how the electrical signal generated by the infinite number of neurons that constitute it, can be acquired through electroencephalography.

Cortical pyramidal neurons are excellent dipoles due to their unique anatomical structure and long apical dendrite perpendicular to the cortical surface. The superficial or deep location of synaptic input determines the direction of such dipole (see figure 30 for a schematic description). Deferring the subtle regulatory mechanisms of activation elsewhere, a superficial excitation causes a negative scalp potential (inhibitory postsynaptic potential IPSP). In contrast, a positive scalp potential (excitatory postsynaptic potential-EPSP) is due to a deep excitation. Both types of postsynaptic potentials are characterised by changes in intra- and extracellular ion fluxes that occur with a precise directionality: mainly Na+ entry from the extracellular space into the cell in the EPSP, K+ exit in the reverse direction with concomitant Cl- entry in the IPSP. Thus, the EEG is the total of all excitatory and inhibitory postsynaptic potentials of cortical pyramidal neurons that produce a vertical dipole perpendicular to the scalp (Kirschstein and Köhling, 2009); practically the same as the sea wave sweeping ashore is made up of the sum of the energies it sums up on its journey. However, as shown in figure 30, the situation at inhibitory synapses is reversed.

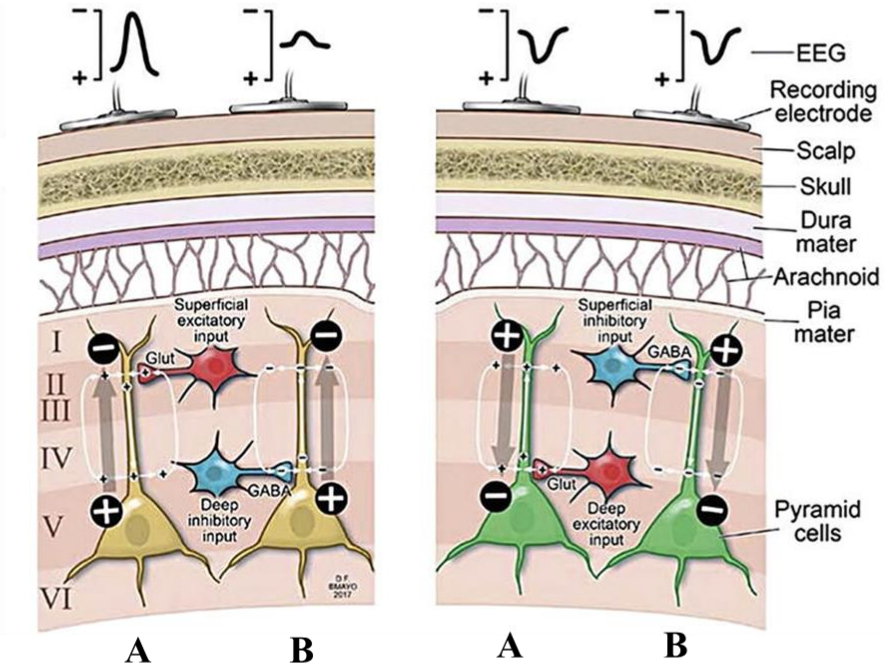

Figure 30. Schematic drawing of the scalp EEG registering negative and positive deflections elicited from summated EPSPs and IPSPs derived from pooled pyramidal cells crossing the six laminar layers of the cerebral cortex. Cells releasing glutamate (B) and GABA (A) provide excitatory and inhibitory superficial and deep synaptic connections resulting in an electrophysiological sink or source. EEG = electroencephalography; EPSPs = excitatory postsynaptic potentials; GABA = gamma-aminobutyric acid. (Source: modified by Tatum et al., 2018).

II. 4. How Postsynaptic Potentials become EEG Waves

Ionic charge shifts caused by EPSP or IPSP generate extracellular potentials, Local Field Potentials (LFPs), which are the essence of the electroencephalographic signal recorded by the scalp. Electroencephalography is a graphic representation of the voltage difference between two cerebral locations plotted over time (Olejniczak, 2006). Therefore, the surface electroencephalogram consists in the recording of the bioelectrical activity from the scalp because of the fluctuation of large populations of cortical neurons; these extracellular current flows are generated by the spatial summation of the postsynaptic potentials of the activated cells (figure 31).

Finally, technically, EEG is the graphical representation of the potential difference between an "active" electrode placed above the seat where the neuronal activity occurs, and the “indifferent” electrode, located at a certain distance from the first. It is a dynamic measure as the potential difference is represented as a function of time. Therefore, the surface EEG measures the electric potential difference between different areas of the scalp and reflects the current flowing in the cerebral cortex during synaptic activation of the dendrites of many pyramidal neurons, which lie just below the surface of the skull. The amplitude of the recorded signal depends on the distance between the two electrodes and grows as the distance between the electrodes increases (Brienza and Mecarelli, 2019) (see figure 31).

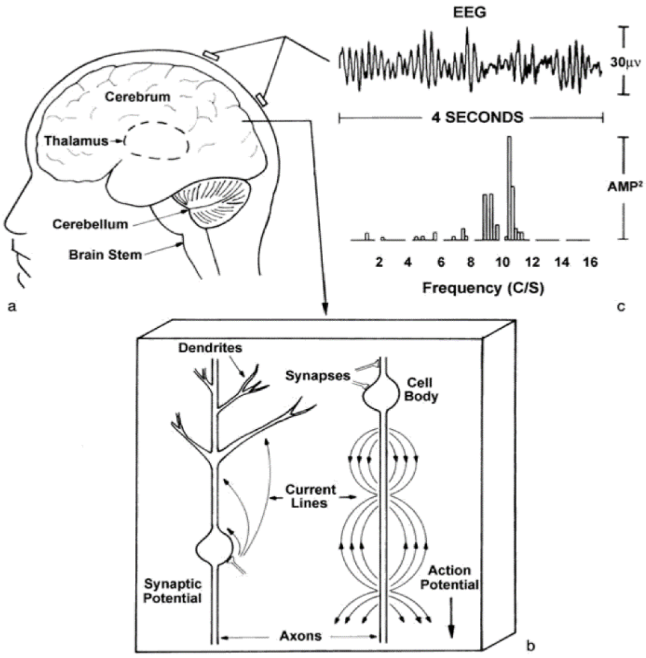

Figure 31. a) A synthetic representation of the human brain. b) Section of the cerebral cortex showing microcurrent sources due to synaptic and action potentials. Neurons are actually much more closely packed then show, about 10^5 neurons per mm^2 of surface. C) Each scalp EEG electrode records space averages over many square centimetres of cortical sources. A four-second epoch of alpha rhythm (which it will be explored in more details later) and its corresponding spectrum are shown. (Source: Nunez and Srinivasan, 2006).

II. 4. 1. EEG Electrodes

Electrodes are the connecting system between the tissues where bioelectric activity is generated, or through which it is conducted, and the amplifier inputs. They can be of different types and shapes, but they are still an interface between the *subject recorded* and the *recording device*. Clearly, without going into detail, the ideal electrode should not distort the signal nor generate significant artifact, however in reality is very difficult to accomplish. Standard recording wet electrodes for scalp are bridge or pad electrodes, cup electrodes and middle electrodes (see Di Flumeri et al., 2019) for a recent comparison between types of electrodes. A cup electrode (or disc electrode, the typology that was used to conduct the empirical research that is the subject of this thesis) is usually composed of a silver/silver chloride (Ag/AgCI) disc plate (that can be considered the gold standard in biopotential detection, (Fiedler et al., 2015), partially concave in its inner part, with a small hole in the middle. In this case, the cup is integrated in a head cap with the wire connecting it to the amplifier and this cable can be directly welded to the plate or detachable (Mecarelli and Panzica, 2019). In fact, although a wide range of choices are available when performing a modern EEG recording, the conventional set up is illustrated in figure 32.

Figure 32. (a) A conventional EEG set up with metal electrodes on the scalp held in place by a n head cap. Long wires connect them to recording instrumentation. (b) Close-up of an electrode making contact with skin via a conductive gel. (Source: Casson et al., 2018).

Once all the electrodes are placed in the cap, and the latter is positioned on the patient's head, it is possible to insert a conductive gel through the hole on the electrodes, to facilitate their contact with the skin, and an impedance check must be carried out, an acceptable value is <20Ω (Nunez and Srinivasan, 2006). All electrodes inserted into the cap are firmly connected to the wires, which in turn are connected to the amplifier through its connector. This method of preparation is usually more comfortable for the task participant's (thinking about children) and it guarantees montages with multiple electrodes achieved in a relatively short time, with good recording reliability, especially in the case of long-term monitoring or sudden movements of the recorded subject (electrodes are not easily moved). Using these caps, however, it is not possible to reduce easily skin-electrode impedances and the electrode placement cannot be modified in case of any particular cranial conformations, skin wounds (Mecarelli and Panzica, 2019) or other artifact generators like the cochlear implants in this specific work. Moreover, and the experience in this case has been teacher having registered at least 200 subjects in the last few years, when the excessive amount of gel is inserted between the skin and the electrode, slow and large sliding artifacts can also occur easily. Anyway, the main benefit of these electrodes is that they guarantee reliable recordings, with flexible and easily adjustable positioning. However, the gel tends to dry over time, and this causes an increase in impedance, moreover the patient can be sensitive to it (both epidermically and psychologically), thus allergic reaction, skin irritation or crying fits can occur (the latter especially in children). Moreover, for long term monitoring, due to change in humidity, the variation in the gel impedance causes artifacts. Furthermore, skin preparation and gel application are time consuming when a high number of electrodes is required (Salvo et al., 2012). However, this method is consistent over time and is more resistant to wire movements and traction, therefore is more suitable for more long-term EEG monitoring as in the experimental protocols elaborated for the researches here presented (see part III of the present work for details).

Ag/AgCl electrodes with wet conductive gel are still almost universally used for biopotential recordings in clinical and research applications being well-characterized and studied over many decades see (Chi et al., 2010) for a methodological review. However, as mentioned, they are not without complications and artefacts due to the use of gel. To overcome these clinical and experimental drawbacks, dry electrodes have been developed in recent years, which do not require the use of conduction gel during the experimental set-up. In the laboratory that I have relied on to develop my experimental work during these exciting years of research, a new signal acquisition device, linked to a European project won by the team of engineers coordinated by Prof. Fabio Babiloni, the results of which have been published (Sciaraffa et al., 2022; Rooseler et al., 2022) and are currently being applied in numerous applications. I had the opportunity to collaborate on the drafting of experimental protocols and the acquisition of EEG data on tinnitus patients using this new methodology. The results of the protocol performed are still under analysis and are not included in this thesis. Likely, they will be further investigated in a post-doctoral phase. Moreover, using this new acquisition device, I am devising a project in the field of musical neuroaesthetics for my post-doctoral work. Here I would like to report how experience of working with different EEG signal acquisition tools has made me even more aware of the principles underlying the study of electroencephalographic signals and the importance of the synergy of interdisciplinary teams, in carrying out clinical and non-clinical studies in auditory cognitive neuroscience.

II. *4. 1. 1. Electrode Placement System*

When Hans Berger recorded the first human EEG, he had only two electrodes, placed in the anterior and posterior regions of the head, believing it to be an efficient way to measure global cortical activity. Later research showed that, in fact, EEG activity varied significantly depending on the area of the scalp from which it was recorded. Once again, it is the human eye that, by observing different rhythms in different brain regions, encouraged the use of more electrodes and more recording channels, and standardisation of positioning soon became necessary to compare results. A committee of the International Federation of Societies for EEG and Clinical Neurophysiology (IFSECN)[12], led by Herbert Jasper (1906-1999), then began work on a specific electrode placement system to be used in all laboratories. The first standardised system, published in 1958, is still universally used today and known as the International System 10-20 (IS 10-20) (Jasper, 1958). The system is called 10-20 because the distances between the electrodes are measured as 10% or 20% of the skull. The letters of the electrodes correspond to their position on the skull (F-frontal, T-temporal, C-central, O-occipital), with odd-numbered electrodes on the left hemisphere and even-numbered electrodes on the right hemisphere. The precise correct positioning of the electrodes on the scalp according to IS 10-20 is achieved by drawing imaginary lines from specified anatomical reference points. The lines are mutually perpendicular and are represented by the anteroposterior sagittal midline, connecting nasion to inion via the vertex; the sagittal midline and

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[12] https://www.ifcn.info/#

the latero-coronal line, from the right preauricular point to the left preauricular point, via the vertex (figure 33).

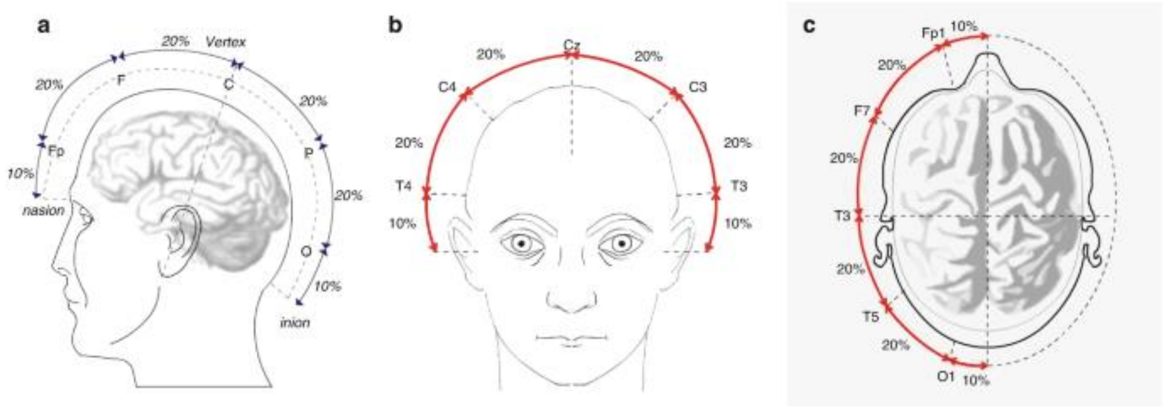

Figure 33. Traditional 10-20 System ideation. (a) Anteroposterior mesial line, connecting nasion and inion; (b) latero-lateral coral line, connecting the two preauricular points through the vertex; (c) sagittal lateral longitudinal line, connecting nasion and inion. (Source: Mecarelli, 2019b).

Along the lines thus drawn, positions were fixed: this measuring system identifies 21 standard electrode positions, including the reference auricular electrodes (A1, A2) (figure 34a). As Mecarelli (2019b) emphasises, the 10-20 system has, in a short time, defined a standard positioning of scalp electrodes, enabling a reliable comparison of data from different laboratories. There are, however, critical issues; for example, the system does not take into account the differences in shapes that human heads can have and that not all of them are symmetrical. Furthermore, the positioning of the electrodes and their number (21) do not allow for more advanced neuroimaging studies to obtain the necessary data to specifically identify the anatomical structures of the brain. Therefore, the development of EEG research (e.g. high-density EEG) and source localisation methods has made it necessary to increase the electrode array. For this reason, a modified 10-20 nomenclature, the 10-10 nomenclature, was proposed and accepted internationally (Seeck et al., 2017; Acharya et al., 2016) (figure 34b).

The modified nomenclature (10-10) provides for the placement of more than 70 electrodes on the scalp, placed along 11 sagittal and 9 coronal chains, with some changes in terminology. Without elaborating further, it is worth noting that the advantage of this new labelling is that all electrodes designated with the same letter are placed in the same coronal line and that all electrodes placed along the same sagittal line have the same postscript number (except Fp2/Fp1 and O2, O1). A further extension of the 10-20 system, called the 10-5 system, with 345 positions, was proposed in 2001, but it has not yet been accepted by ACNS and IFCN (Oostenveld and Praamstra, 2001).

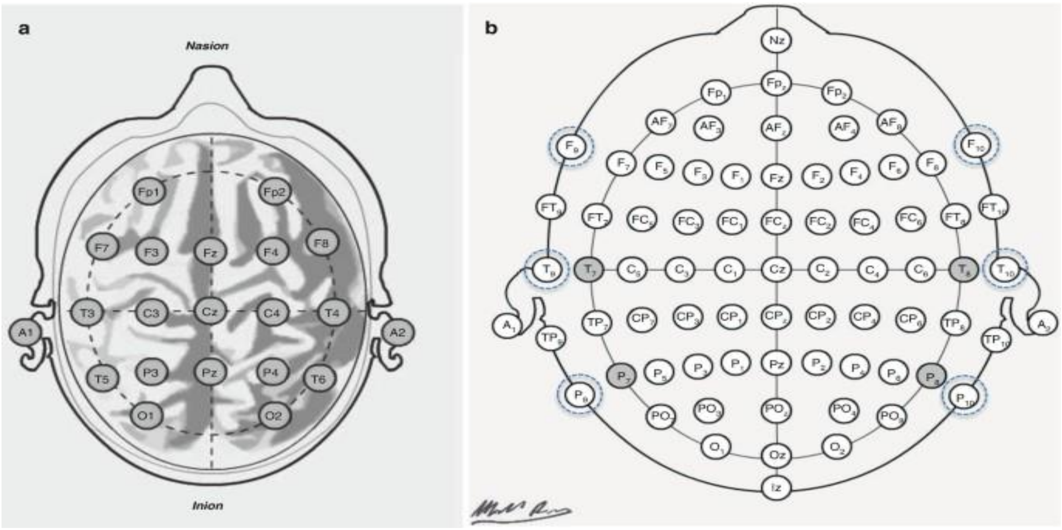

Figure 34. (a) Standard position of the 19 scalp electrodes and of the 2 ear reference electrodes, according to traditional 10-20 system; (b) modified 10-10 system, with 73 electrodes positions on the scalp and 2 ear reference electrodes. (Source: Mecarelli, 2019b).

Finally, it was considered important to partly investigate here the system by which the electrodes are placed on the scalp, as it is from their positioning that the data on which the main experimental questions and subsequent investigations are based originate. During the research years for the PhD, I had the opportunity to conduct experiments using only frontal electrodes (see experimental research E), 20 electrodes (experimental work A, B, C, D) and up to 25 electrodes used for a recent data acquisition for the WHEMORCHING[13] research project with analysis still in progress. Cardinal points are essential for navigating adventurous routes for even the most experienced navigator (researcher).

II. *4. 1. 2. EEG Signal Analysis and its Phases*

Thus, eventually we arrive to the analysis of the signal acquired on the cerebral scalp.

We started this general part of the work from the auditory signal that propagates in the air and is picked up and processed by the auditory system to arrive, going up the anatomical pathways, to trigger processes at the cortical level and how the signals manifest themselves at the level of the scalp. Although it is not possible here to provide all the details on the electroencephalographic signal and its analysis (which are clearly provided in the materials and methods session of the studies in the empirical part of the thesis), we will now briefly describe the various stages of processing that allow the signal to be processed and data to be obtained with which to analyse the brain waves in more detail. So, the EEG framework with frequently used modules in most of the EEG analysis is shown in figure 35.

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[13] Neurophysiological CHaracteristics of an EMOtional auditory/visual WoRkING memory task in Children with and without Hearing loss

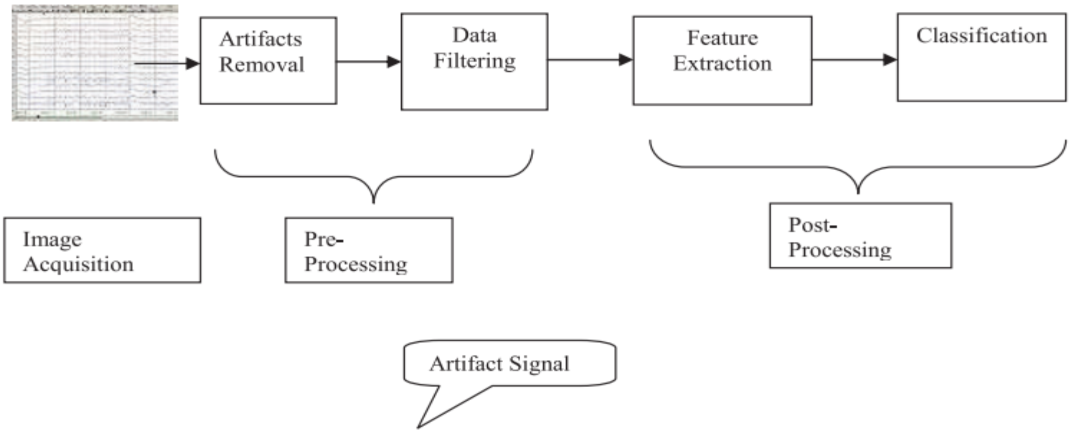

Figure 35. EEG signal processing principal phases. (Source: modified by Kumar and Bhuvaneswari, 2012).

In EEG Signal *Image Acquisition* phase, raw EEG signals will be collected directly from the scalp of the brain. Second phase is *Pre-Processing* stage which consist in two processes such as, *Artifacts Removal* and *Data Filtering*. Identification and removal of artifacts, undesired signals that may introduce changes in the measurements and affect the signal of interest (Urigüen and Garcia-Zapirain, 2015) is a challenging phase in signal acquisition and analysis. Artifacts may be triggered due to various factors such as, had motion during acquisition, physical problems in electrodes, and connectivity issues between head and device. These artifacts will also create signals with abnormal frequency and shapes.
It is possible to remove motion interference algorithmically. Most approaches for removing such interference, and recovering clean EEG underneath artefacts, are based upon signal decomposition techniques such as independent component analysis (ICA) and principal component analysis (PCA) (Cohen, 2014). ICA is particularly popular and implemented in the widely used EEGLab toolbox for MATLAB (Natick, 2022), (Delorme and Makeig, 2004), which allows artefact removal to be performed without having to be involved with the full mathematical details. Artifact removal completed, next step is *Feature Extraction* where feature of the signal can be derived using various signal processing technique like for example Fourier Transform, Wavelet Transform for a comparison between the two see (Akin, 2002). Fourier transform is the method that has been used for the protocol object of this work, allows the raw EEG signal to be classified into 4 bands: Gamma, Delta, Beta, Alpha, Theta. This classification -as seen- is historical and based on signal frequency. Some waves are recognized based on their shape, head distribution and asymmetry property (Salmelin and Hari, 1994). The classical frequency bands of EEG are defined as follows and schematically represented in the figure 36. Some possible functionalities of the bands, observed in the extensive research over the years, are also reported, given that with the use of such technology has been possible over the years to detect the involvement of circumscribed cortical areas of the two hemispheres related to sensory and motor events see for example (Gevins et al., 1989; Nunez and Cutillo, 1995; Urbano et al., 1998; Babiloni et al., 1997; Babiloni et al.,

2007b; Borghini et al., 2014). However, despite numerous observations, the precise functional significance of cortical rhythms is still unknown (Purves et al., 2013b). Indeed, the reason for the relevant oscillatory activity in the brain is an enigma that has engaged electrophysiologists, neurobiologists, and neuroscientists for more than seventy years. This enigma prompts even more in-depth research into brain waves.

- *Delta* (δ): 0.1 to <4 Hz. The rhythm is associated with non-REM deep sleep states with a frequency that can be associated with pathological situations. It was first described by Walter in 1936 as an EEG marker for cerebral tumour.
- *Theta* (θ): 4-8 Hz, should be divided in Theta 1 (slower:4-6 Hz) and Theta 2 (faster: 6-8). Theta band can be associated (as shown in the empirical part of this work) with particular emotional states or mental processes of problem solving and memory.
- *Alpha* (α): 8-13 Hz) is proper for an EEG tracing register in adults during quiet wakefulness with eyes closed. Even within this frequency band, it is possible to distinguish slow Alpha (8-9 Hz), intermediate (9-11 Hz) and fast Alpha (11-13 Hz).
- *Beta* (β): 13-30 Hz; this band is related to the mental and cognitive process of various types, anxiety and state of alert. It is also induced or increased by drowsiness and light sleep.
- *Gamma* (γ) 30-80 Hz: is associated with high performance (physical and intellectual) and deep concentration.

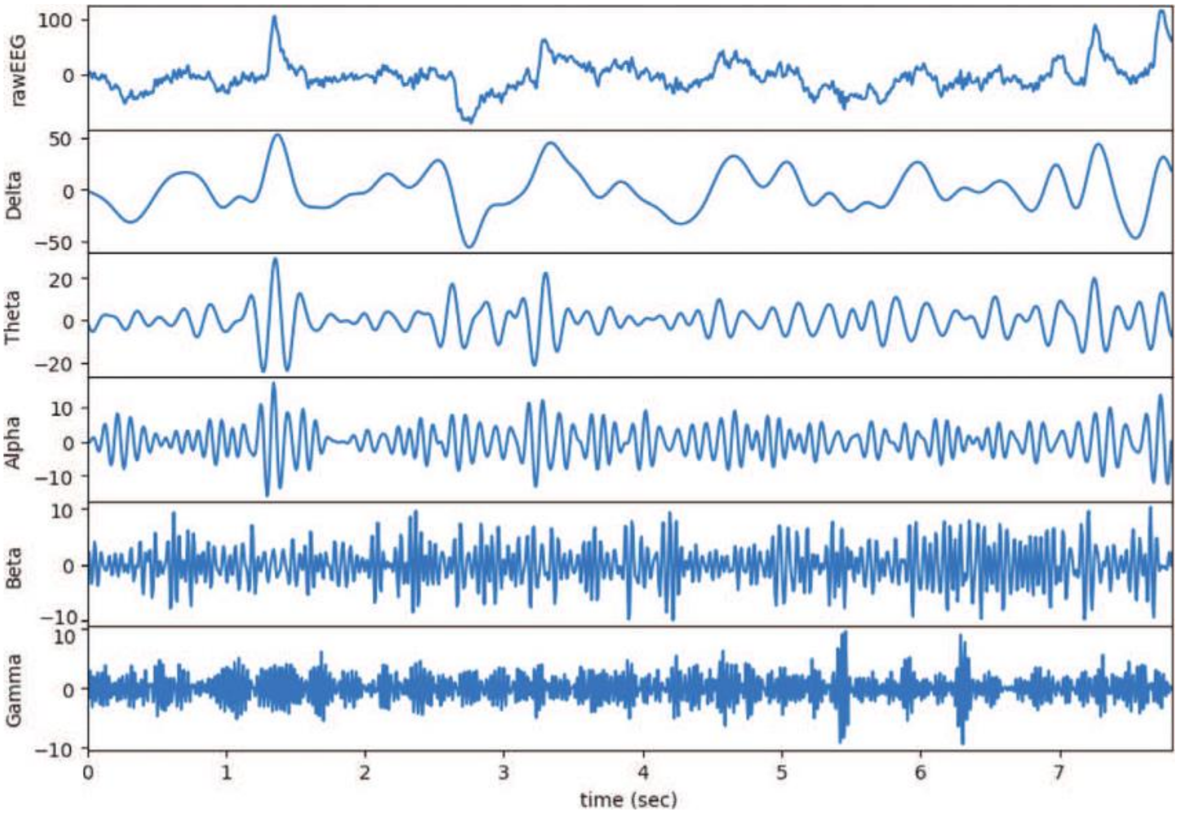

Figure 36. The signal channel raw EEG signal and corresponding frequency bands: Delta (0- 4 Hz), theta (4 -8 Hz), alpha (8-13 Hz), Beta (13-30 Hz), gamma (>30 Hz). (Source: Bajaj, 2020).

Given the breadth of studies in clinical and experimental settings that use EEG analysis, the interested reader is referred to the empirical studies presented in the second part of this work. Finally, as mentioned earlier, it is important here to emphasise that although research has made great strides in the application of EEG to the analysis of human behaviour in both adults (Feyissa and Tatum, 2019) and children (Bell and Cuevas, 2012) numerous questions and controversies remain unresolved concerning the nature and specific functions of the different frequency bands. Especially in children and young adults, when assessing the EEG, I realised how important it is to follow a multi-level approach, thus combining the assessment of the neurophysiological component with the behavioural and psychological one. Thus, EEG research is placed within an integrative framework that recognises the two-way interaction between the brain and behaviour (Saby and Marshall, 2012); see (Miller, 2010) for an overview on relationship between psychology and biology.

III. EXPERIMENTAL RESEARCH

Introductory considerations and summary of the scientific questions

The basic purpose of my work has been the scientific understanding of cognitive and emotional processes. The main technological tool which I demonstrated to be useful at this scope, has been the EEG and in particular its quantitative suitability in successfully detection of brain cognitive and emotional mechanisms.

Notably, the outcomes of the WM could be analysed and studied in different populations. I mostly concentrated on children population comparing normal hearing and cochlear implants users. This has been a complete work including the experimental definition of a normal hearing verbal WM benchmark (**A**).
A significant comparison could be carried between NH and unilateral cochlear implant (CI) users (**B**). The listening with unilateral CI in children was also electrophysiologically investigated during emotions processing of different emotional vocal auditory stimuli (**C**). Moreover, pleasantness and effort when listening to music in noise was investigated through EEG in groups of adults with unilateral cochlear implants (**D**). Of course, the complexity of this matter rises from the occurrence of a large variety of emotive inputs and here we report the study on the emotional reaction to stimuli conditioned by other clinical pathologies such as smoking addiction (**E**).

The multidisciplinary complexity of this research field originates also from the different abilities of attention and its variability detected through psychometric assessment batteries in cochlear implant users both children (**F**) and adults (**G**). These two latest investigations (**F** and **G**) have given me the cognitive basis to explore attention through neurophysiological methods in the future.

During the pandemic period we could not perform laboratory experimental studies because we have to avoid the contact with people, however we could perform studies through remote tools (as online survey and facial coding online detection). This has allowed me to broaden the landscapes also on the investigation of the effectiveness of distance learning in students with and without hearing difficulties (**H**). This has been a contribution to a more general debate on the psychological and neuropsychiatric effects of distance experienced in pandemic (**I**). Finally, the emotional reaction of university students to artistic product such as poetry, has been the object of the last scientific question elaborated during pandemic (**L**).

So, **A**, **B**, **C**, **D**, **E**, **F**, **G**, **H**, **I**, **L**, refer to the different scientific questions addressed with results published in peer reviewed journals as reported in the following.

A. “Is there a cross-modality in the neurophysiological processing of auditory verbal and visual working memory in children? If so, is it possible to identify standard patterns of neurophysiological activation during verbal WM activities independently of the sensory modality of the stimuli processed?”.

- “Neurophysiological verbal Working Memory patterns in children: searching for a benchmark of modality differences in audio/video stimuli processing”.

B. “What is the impact of unilateral cochlear implant use in children with hearing loss on the neurophysiological patterns of auditory and verbal working memory processing? Is it possible to identify a sensory-related neurophysiological pattern of verbal working memory in this clinical population?”.

- “Gamma-Band Modulation in Parietal Area as the Electroencephalographic Signature for Performance in Auditory–Verbal Working Memory: An Exploratory Pilot Study in Hearing and Unilateral Cochlear Implant Children”.

C. “Are the emotional auditory and visual stimuli processed in a similar way in cochlear implant user than NH children? Is it possible to identify a brain lateralization EEG patterns in the processing of vocal auditory emotional stimuli?”.

- “Higher right hemisphere gamma band lateralization and suggestion of a sensitive period for vocal auditory emotional stimuli recognition in unilateral cochlear implant children: An EEG study”.

D. “Is it possible to investigate pleasantness and effort in the perception of classical music under different listening conditions in adult CI users through neurophysiological indices and cognitive-behavioural measures?”.

- “Musical effort’ and ‘musical pleasantness’: a pilot study on the neurophysiological correlates of classical music listening in adults normal hearing and unilateral cochlear implant users”.

E. “Is it possible to investigate through electroencephalography the unconscious emotional responses to stimuli related to tobacco addiction in smokers?”.

- “Smoke signals: A study of the neurophysiological reaction of smokers and non-smokers to smoking cues inserted into antismoking public service announcements”.

F. “What is the specific contribution of auditory selective attention on linguistic performance in children with cochlear implants?”.

- “The influence of auditory selective attention on linguistic outcomes in deaf and hard of hearing children with cochlear implants”.

G. "Is it possible to identify the effects of specific cognitive abilities such as working memory and attention on speech perception in elderly cochlear implant users in order to obtain a cognitive target for intervention to support the patient?".

● "Neuropsychological Functions and Audiological Findings in Elderly Cochlear Implant Users: The Role of Attention in Postoperative Performance".

H. "What are the psychological characteristics of online learning during the Covid-19 lockdown on Italian students with and without hearing loss and their parents forced to isolate themselves and distance themselves from the world?" Moreover, did online learning impact students with hearing loss and their families more than their peers without?".

● "School wellbeing and psychological characteristics of online learning in families of children with and without hearing loss during the Covid-19 pandemic".

I. “An insight into online education during pandemic isolation, reflection on student experiences and school inclusiveness”.

- “Come (si) sentono i ragazzi? Riflessioni su Didattica a Distanza e Sordità ai tempi della pandemia”.

L. “How does the emotional response to poetic stimuli vary according to their mode of enjoyment in university students with scientific and literary backgrounds and according to the presence of alexithymia during the social distancing imposed by the covid 19 pandemic?”.

- “Poetry in Pandemic: A Multimodal Neuroaesthetic Study on the Emotional Reaction to the Divina Commedia Poem”.

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Research Article

Neurophysiological Verbal Working Memory Patterns in Children: Searching for a Benchmark of Modality Differences in Audio/Video Stimuli Processing

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Exploration of specific brain areas involved in verbal working memory (VWM) is a powerful but not widely used tool for the study of different sensory modalities, especially in children. In this study, for the first time, we used electroencephalography (EEG) to investigate neurophysiological similarities and differences in response to the same verbal stimuli, expressed in the auditory and visual modality during the n-back task with varying memory load in children. Since VWM plays an important role in learning ability, we wanted to investigate whether children elaborated the verbal input from auditory and visual stimuli through the same neural patterns and if performance varies depending on the sensory modality. Performance in terms of reaction times was better in visual than auditory modality ($p = 0.008$) and worse as memory load increased regardless of the modality ($p < 0.001$). EEG activation was proportionally influenced by task level and was evidenced in theta band over the prefrontal cortex ($p = 0.021$), along the midline ($p = 0.003$), and on the left hemisphere ($p = 0.003$). Differences in the effects of the two modalities were seen only in gamma band in the parietal cortices ($p = 0.009$). The values of a brainwave-based engagement index, innovatively used here to test children in a dual-modality VWM paradigm, varied depending on n-back task level ($p = 0.001$) and negatively correlated ($p = 0.002$) with performance, suggesting its computational effectiveness in detecting changes in mental state during memory tasks involving children. Overall, our findings suggest that auditory and visual VWM involved the same brain cortical areas (frontal, parietal, occipital, and midline) and that the significant differences in cortical activation in theta band were more related to memory load than sensory modality, suggesting that VWM function in the child's brain involves a cross-modal processing pattern.

1. Introduction

The term working memory (WM) [1] refers to the type of memory that is active and relevant for short periods of time, usually only seconds [2]. Specifically, it is the theoretical construct used in cognitive neurosciences to refer to the system or mechanism underlying the maintenance of relevant information during cognitive task performances [3, 4]. Baddeley-Hitch's WM model proposes a tripartite system organized in a central executive and two subsidiary systems: the phonological loop, capable of holding verbal information, and the visuospatial sketchpad, which exercises a parallel function for spatial information [1, 5].

Although the WM multicomponent model is influential in scientific thinking, its neural basis remains poorly specified [6].

There is evidence that WM provides a mental workspace used in many fundamental learning activities during lifespan, including *literacy* [7, 8], *reading* [9], and *numeracy* [10]. These findings have important implications in education, particularly for children with neurodevelopmental disorders and sensory deficits [11].

N-back task [12] has become a prototypical measure in functional neuroimaging studies that allows identification of the neural mechanisms supporting WM [13] (see [14] for a meta-analysis). In fact, studies consistently find that n-back performance is associated with activation in prefrontal and parietal cortical regions widely recognized as the primary neural substrates that underlie working memory processes [2, 14–18] and in particular visual and auditory stimuli processing [19, 20]. Moreover, patterns of neural activation associated with n-back performance have been shown to vary with the type of information held in working memory (e.g., verbal or spatial), as well as task difficulty (i.e., 0-, 1-, and 2-back) (see [21, 22] for review).

It has been suggested that the prefrontal cortex (PFC) is critical for resilient information maintenance during WM tasks [23]. Given its functional connections with the posterior parietal cortex, Dorsolateral PFC plays a crucial role in both verbal and visuospatial WM [14, 24] (see [25] for review). Moreover, stronger frontoparietal synaptic connectivity may be one of the mechanisms involved in WM capacity development during childhood [26]. Investigators have mapped WM-related activity to sensory association cortices and PFC and some regions show specificity to sensory stimuli modality (see [27, 28] for review).

Adults have been demonstrated to have functional hemispheric specialization for WM with a refinement of verbal processing operated by the left hemisphere, whereas the right hemisphere appears more specialized in visuospatial processing [22, 29–31]. Few studies have examined this potential sensorial input dissociation or whether more distinct or lateralized patterns of brain responses and considered signature of WM emerge across development [32]. In fact, brain structures and neural processes subserving WM continue to develop during childhood [33, 34], and it is known that changes in PFC are related to cognitive development achievements occurring during childhood as well [35–37].

Neuroimaging assessments for verbal [38] and visuospatial [39–41] stimuli support the evidence that WM-related activation is greater and more widely distributed in the child brain than the adult brain [33, 42]. This may reflect ongoing maturation and synaptic fine tuning during development [37].

EEG neuroimaging studies, principally in adult populations, have evidenced enhanced activity during WM load-specific modulations by different bands, in particular theta, alpha, and gamma in numerous brain areas [43–47]. Moreover, a brainwave-based mental engagement index (EI) previously defined by Pope and colleagues [48] as

$$\mathrm{EI} = \frac{\mathrm{Beta}}{\mathrm{Alpha} + \mathrm{Theta}}, \qquad (1)$$

has proven to be successful in distinguishing brain attentive states and to correlate with emotions and mental workload in memory tasks [49–52]. Furthermore, McMahan and colleagues [53], comparing EI with other EEG engagement indexes (frontal theta, ratio of frontal theta to parietal alpha), found the ratio between beta and the sum of alpha and theta to be the best algorithm for calculating the engagement levels of players playing video games. Research has shown that classifiers using physiological features are able to determine the level of cognitive activity in tasks with a high level of accuracy [54]. However, studies that have applied EI to assess children's cognitive engagement are rare (see [55, 56]), and to the best of our knowledge, there are no published EEG studies involving the assessment of WM through the n-back task in children younger than 13.

To date, there have not been many investigations on the specific relationship between cognitive and neurophysiological developmental changes in WM functions during childhood [57–60]. Furthermore, several studies focus on the visuospatial component of WM processing (e.g., [61–63]). Better understanding of the development of WM functions would help in the determination of what is normal and what is pathological at different ages and in the development of new learning, teaching, and cognitive training strategies [64–69].

Verbal WM (VWM) is a specific human form of WM that appears to play a significant role in language comprehension and problem solving [70]. It is particularly important given the role that linguistic processes play in the higher-cognitive processes [18]. Most of our knowledge of the neural network underlying VWM is based on studies using visually presented stimuli [22, 39, 71]. There have been few reports of investigations on the purely neural basis of auditory VWM [72–74], and even fewer have directly examined modality differences using similar tasks in a within-subjects design [6]. Specifically, with the aim of digging into the neural mechanisms underlying the model of processing of the verbal components of WM (e.g., phonological loop [75]), only four neuroimaging studies concerning adult populations have considered the effect of n-back task modality on brain activation [6, 76–78]. Those studies reported contrasting findings and employed neuroimaging techniques different from EEG, which we selected for its high temporal resolution. It is noteworthy that most of the published studies on working memory involve the use of brain-imaging techniques that are more invasive and less ecological than EEG, such as fMRI (e.g., [63, 79–81]) and PET (e.g., [82]) that are often impractical for use on children [59, 83]. Precisely, no studies have used EEG to assess neural responses during verbal n-back tasks with different sensory stimuli (visual and auditory), in particular in healthy children.

The aim of this study is to examine EEG activation during VWM processing of *auditory* and *visual* stimuli presented to children during n-back task performance [12].

The distinction between auditory VWM and visual VWM is important, with implications for both theoretical and experimental research on the neural processes underlying WM. In fact, as Crottaz-Herbette and colleagues [6] pointed out, elucidation of similarities and differences in the processing of different types of stimuli can provide insight into the internal representations of stimuli in WM. Indeed, to date, the considerable theoretical debate on WM features is evident in the many cognitive studies (e.g., [84–86]) that investigate whether WM storage is mediated by distinct subsystems for auditory and visual stimuli [5] or by a single central capacity system [87].

Moreover, the discrepancies in the studies regarding adults reported above, on the assumption of an a-modal VWM system [6, 76–78] and the absence of studies on healthy and clinical child populations, evidence a scientific void that must be filled. In an attempt to tackle this issue, the experimental investigation of the neural underpinnings of auditory and visual stimuli processing and the consideration of a possible cross-modal activation during childhood in a VWM task appears extremely important for the evaluation of healthy development of children with or without sensory impairment.

We hypothesized that, in children, the involvement of the theoretical phonological loop [75], which underpins verbal WM processing, is neurally mediated indifferently by both auditory and visual stimuli. Thus, with both visual and auditory-verbal WM n-back tasks, we expected to find the following:

(1) There are no differences in EEG activation patterns in the cortical areas involved in VWM function in response to the two different sensory stimuli.

(2) As largely confirmed by the literature on adults presented above, significant differences in EEG activation in response to auditory and visual stimuli depend only on memory load variations (0-1-2-back) and the EEG findings correlate with behavioral results also in children.

Confirmation of our hypotheses could indicate the possibility of identifying a neurophysiological benchmark of auditory-visual VWM in healthy young individuals, also allowing further comparisons with clinical research groups.

2. Material and Methods

2.1. Participants. Thirteen right-handed children aged 7–13 years (for age and sample size definition (children were selected according to previous studies), see [88, 89]) were enrolled in the study. Two participants were subsequently excluded because of the lack of cooperation in the task training accomplishment. Therefore, the final experimental sample was composed of 11 children (6 M and 5 F; mean age = 10.83 ± 1.87 yr).

Prior to the experiment, participants and their parents were informed about the study. We obtained informed written consent from the parents and verbal assent from the children. Participation in the study was voluntary; participants did not receive compensation for taking part. The experiment was conducted according to the principles outlined in the Helsinki Declaration of 1975, revised in 2000, and approved by the Institutional Ethics Committee of Policlinico Umberto I- Rome (no. 259/2020).

Subject selection was based on diagnostic screening using the *Peabody Picture Vocabulary Test-III* [90], a standardized measure of receptive oral vocabulary, and *Raven's Standard Progressive Matrices* [91], a standardized test of nonverbal spatial reasoning. Both tests use standardized scoring based on participant age ($\mu = 100$, $\mathrm{SD} = 15$). Exclusion criteria for enrollment in the study were left-handed children, due to past evidence of handedness influence on cerebral laterality [92]; children with scores below the standard average for their age (taken from test norms) on PPVT and RPM; and those diagnosed with neuropsychiatric disorders and/or sensorial deficits.

2.2. Experimental Design and Procedure. Participants performed two verbal n-back tasks [12] with varying memory load from 0-back to 2-back during EEG recording: (i) an auditory n-back task (AUD-task) in which stimuli were presented aurally and (ii) a visual n-back task (VIS-task) in which stimuli were presented visually on a computer screen.

Task administration order was randomized across participants. Therefore, approximately half of the participants started with the AUD-task and the second half with VIS-task. In addition, the order of presentation of the n-back blocks was randomized across participants; in other words, it did not follow an increasing level order.

> *Stimuli*: verbal material consisted of auditory and visual stimuli referring to seven consonants (c, g, k, p, q, t, and v), already used and described in previous studies [93–96]. Vowels were excluded in order to reduce the likeliness of participants developing chunking strategies, as suggested in Grimes et al. [97]. Stimuli exposure pretest was performed to ensure correct perception by participants. Auditory stimuli, lasting 500 ms and presented with an interstimulus interval (ISI) of 2500 ms [96], were spoken by a female voice set at 65 dB SPL intensity, in order to ensure comfortable audibility, transmitted by two audio speakers placed at face level 1 meter in front of the participant. Visual stimuli (duration 500 ms; ISI 3000 ms) [88] consisted of the same seven consonants (Consolas font-130) presented one at a time on a grey background in the center of a monitor screen placed at eye level, 50 cm distant from the participant.
>
> Task execution: participants had to respond in the ISI just after the presentation of each letter by pressing a button (D/K) to indicate whether the letter was a target (K) or a nontarget (D); thus, there was a behavioral response in either case. In the 0-back condition, the letter X was the target. In the 1-back condition, a letter was a target when it was the same as the one presented immediately before. In the 2-back condition, a letter was a target when it was the same as the one that presented two letters before. Participants were given

detailed instructions for proper task performance and a training session before the effective measurement session in order to familiarize them with the experimental procedures (Figure 1).

Task structure: the three WM load levels (0-1-2-back) were presented in six blocks (2 for each level) for each task (auditory and visual). The blocks were constituted by 21 randomized stimuli (30% target) [88]. At the beginning of each modality task, there was a *Baseline phase*, during which subjects were asked to remain relaxed, with no task except to look at the screen while auditory or visual stimuli were presented. During the *Baseline phase,* the 7 stimuli were repeated randomly 3 times (duration 500 ms with 3000 ms ISI), creating a 21 item block, analogous to the experimental blocks. Subsequently, the *Task phase* consisted of two randomized presentations of each of the three blocks, which began. Thus, every single session consisted of 3 n-back levels per 2 presentations, for a total of 6 blocks in randomized order for both audio and video tasks (Figure 2). Half of the participants started with the visual stimuli task and the other half with the auditory task.

A Lenovo PC (monitor resolution 1024 × 768) displayed and controlled stimuli presentation and participant responses (reaction times (RTs); correct responses (CRs)) through the software package E-Prime (Psychology Software Tools, Pittsburgh, Pa, Version 3.0).

Procedure: the participant was seated on a comfortable chair in an audiometric test room, and the experimental procedure was explained. In order to reduce muscular artifacts in the EEG signal, participants were instructed to assume a comfortable position and to avoid unnecessary movement. After each *Task phase*, participants indicated the perceived task difficulty (easy-medium-hard) on a stylized image (Figure 3). At the end of the entire experimental session, they were asked to evaluate which of the two tasks (visual or auditory) was the most difficult.

2.3. Behavioral Data Analysis.

Performance was assessed in terms of accuracy (ACC) and RTs. ACC was calculated as the percentage of CRs for each task condition (each n-back level for both auditory and visual modality tasks); RTs were measured from the time of stimulus offset. In order to integrate these two aspects of performance, Inverse Efficiency Score ($\mathrm{IES} = \mathrm{RT}/1 - \mathrm{PE}$) [98] was calculated, where RT is the subject's average RTs for correct answers (target/nontarget), and PE is the subject's proportion of errors for each condition. IES can be interpreted as the RT corrected for the number of errors committed [99].

2.4. EEG Recording and Data Analysis.

EEG was recorded through a digital ambulatory monitoring system (BePlus System -EBNeuro, S.p.A., Italy) with a sampling frequency of 256 Hz. Twenty channels (Fpz, Fz, F3, F4, F7, F8, Cz, C3, C4, T7, T8, Pz, P3, P4, P7, P8, Cp5, Cp6, O1, and O2) were referred to the participants' earlobes, and impedance was kept below 10 kΩ. A 50 Hz notch filter was then applied to remove power interference. EEG signal was band-pass filtered with a 5th order Butterworth band-pass filter (1–45 Hz) to reject continuous components and high-frequency interferences like muscular artifacts. The Fpz channel was used to remove eye-blink contributions by the REBLINCA algorithm [100, 101] without losing data. Other artifacts were eliminated by specific procedures of the EEGLAB toolbox [102].

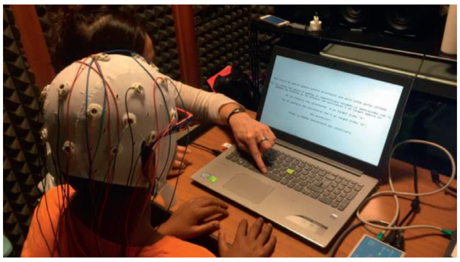


Figure 1: Explanation of task execution before training section. The picture shows a representation of the detailed explanation of the task to each participant before the effective measurement session.

EEG dataset was segmented into epochs starting 500 ms before stimulus onset and ending 2500 ms after its offset. This temporal windowing was chosen to respect EEG stationarity and allow for a high number of observations compared to the number of variables considered in the analysis [103]. Three criteria were applied in order to identify artifacts according to published procedures [55, 104]. In particular, all the epochs exceeding the *threshold criterion* ($\pm 80\,\mu\mathrm{V}$) were marked as artifacts, as well as those that did not meet the *trend estimation criterion (*slope higher than $40\,\mu\mathrm{V/s}$ or less than $0.3\,\mu\mathrm{V/s}$). The EEG epoch was also considered an artifact if the signal sample to sample difference (*sample to sample criterion)* in terms of absolute amplitude was higher than 30 mV, that is, when an abrupt variation (nonphysiological) occurred. Finally, epochs marked as artifact were removed from the EEG dataset such that all analyses were based on clean EEG signals [105–109]. Definition of EEG bands of interest involved the identification of subjective differences in terms of brain activity. Individual Alpha Frequency (IAF) in Hertz was computed on a 60-second long-closed eyes segment, recorded before the *Baseline phase* [110].

Each band was then defined as $\mathrm{IAF} \pm x$ where x was an integer in the frequency domain [110]; thus, electrophysiological activity was divided by filtering EEG signals in the following frequency bands: theta ($\mathrm{IAF} - 6 \div \mathrm{IAF} - 2$ Hz), low alpha ($\mathrm{IAF} - 2 \div \mathrm{IAF}$ Hz); upper alpha ($\mathrm{IAF} \div \mathrm{IAF} + 2$), alpha ($\mathrm{IAF} - 2 \div \mathrm{IAF} + 2$ Hz), beta ($\mathrm{IAF} + 2 \div \mathrm{IAF} + 16$ Hz), and gamma ($\mathrm{IAF} + 16 \div \mathrm{IAF} + 30$ Hz).

Then, the Power Spectral Density (PSD) [111] was calculated for each epoch and channel, using a Hanning window of 1 sec and an overlap of 500 ms. Cortical distribution of band modulation analysis was based on averages of the data for frontal, parietal, occipital, midline, and

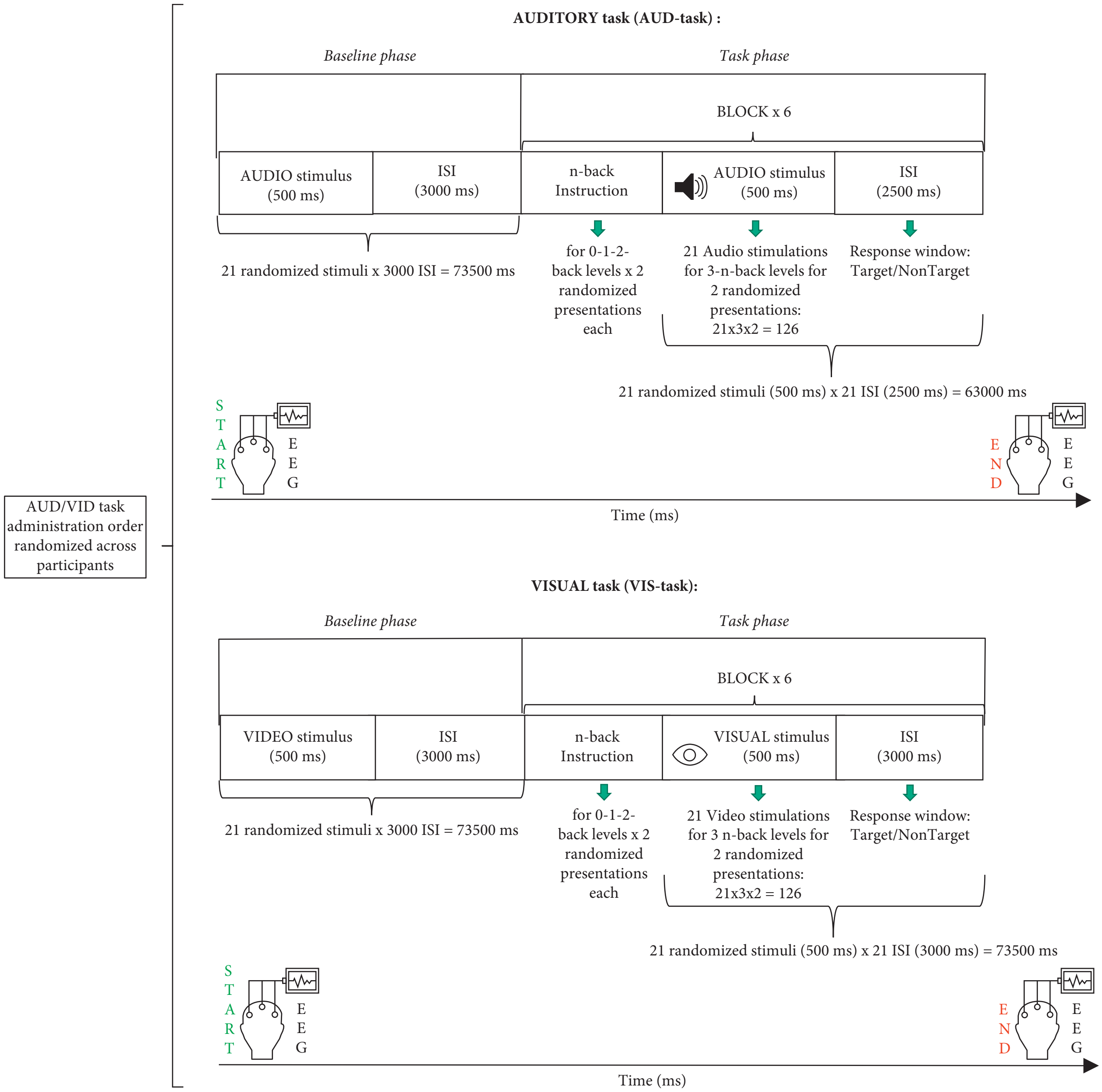


FIGURE 2: Experimental design with trial timeline. Schematic illustration for each of the two n-back tasks (auditory and visual modalities) performed by subjects during electroencephalography (EEG) recording. Each modality task started with the Baseline phase followed by the Task phase.

FIGURE 3: Illustration of perceived difficulty. Each participant was asked to indicate through this image a level of perceived difficulty after each visual and auditory task. *Note*. Translation of the Italian text: *Come ti è sembrato il gioco?* = How was the game?; *Facile* = easy; *Medio* = medium; *Difficile* = hard.

hemisphere electrode locations. The specific channels considered were frontal, F3, F4, F7, F8, and Fz; parietal, P4, P3, P7, and P8; occipital, O1 and O2; midline, Fz, Cz, and Pz; left hemisphere, F3, C3, T7, P3, and O1; and right hemisphere, F4, C4, T4, P4, and O2.

Moreover, EI [48] was calculated according to the formula specified above.

PSD data were normalized with respect to the baseline to limit influences on scores due to subjective stimuli perception on VWM EEG recording [112].

2.5. Statistical Analysis. The statistical analyses were conducted for neurophysiological and behavioral data, respectively. The Shapiro–Wilk normality test [113] was applied to the datasets under investigation. Then, depending on the results, parametric analysis of variance (ANOVA) or nonparametric ANOVA [114] was done. Both behavioral and neurophysiological values were entered in a 2×3 factorial ANOVA with 2 factors: factor modality (with two levels: *audio* and *video*) and factor load (with three levels: *0-1-2*). Duncan's post hoc test [115] was used to investigate statistically significant results of ANOVA tests; partial eta squared (η_p^2) effect sizes [116, 117] were reported. Finally, Pearson's correlation coefficient (r) [118] was used to assess the relationship between behavioral data and neurophysiological values. An alpha value (α) of 0.05 was used as the cutoff of significance [119].

3. Results

3.1. Behavioral Results. Behavioral results (RT, ACC, and IES) are presented in Tables 1 and 2.

RTs and IES increased, and ACC decreased with increasing memory load. Post hoc analysis showed that auditory modality produced significantly longer RTs than visual modality ($p = 0.008$) (Figure 4(b)) and showed a significant increase in RTs as the n-back level increased ($p < 0.001$) both between 0- and 2-back and between 1- and 2-back (Figure 4(a)) regardless of modality.

The overall ACC score percentages were greater during all auditory n-back levels (96.96, 87.60, and 87.01 for 0-1-2 levels, resp.) than for visual ones (90.90, 83.33, and 74.02 for 0-1-2 levels, resp.). Post hoc analysis showed significantly lower accuracy for both the 1-back and the 2-back compared to the 0-back level ($p = 0.007$ and $p < 0.001$, resp.), independently of modality (Figure 4(c)). This trend was reflected in the IES data: post hoc results showed significant differences between 0 and 2 and 1 load ($p < 0.001$ and $p = 0.028$ resp.) and between 1 and 2 loads ($p = 0.002$) (Figure 4(d)).

3.2. Neurophysiological Results. EEG activation during session recordings is shown in Figure 5; neurophysiological statistical results are shown in Table 3.

Post hoc analysis regarding the frontal area revealed that theta power was higher in 2-back than in 0-back level tasks ($p = 0.008$) (Figure 6(a)). Similar increased theta band activity was also evidenced in the midline area, where post hoc analysis showed higher activation related to increasing n-back task difficulty (from 0-back to 1-back levels, $p = 0.013$; from 0-back to 2-back levels, $p = 0.001$) (Figure 6(b)).

The remarkable effect of load factor on theta band activity was also evidenced in the left hemisphere comparing 2- and 0-back levels ($p = 0.001$) by post hoc analysis (Figure 6(c)). The significant different activation on theta band did not depend on modality factor. Differently, post hoc analysis of gamma activity in the parietal area showed sensitivity to audio modality for each level of load × modality interaction, except for 0-back video condition ($p = 0.165$) (Figure 7). Specifically, gamma activation was lower during the 2-back level in the audio task than for the same level in the video task ($p = 0.017$) and also compared to 1-back level audio ($p = 0.005$) and video ($p = 0.005$) stimulation. The difference in gamma activation during the audio 2-back level condition was even more pronounced when compared to that seen with the 0-back level in audio presentation ($p = 0.004$).

Finally, the post hoc test evidenced increased EI values comparing both the 2-back and 0-back levels ($p < 0.001$) and the 2-back and 1-back levels ($p = 0.031$), regardless of modality. Moreover, a negative correlation was observed between EI values and reaction times ($r(64) = -0.36$; $p = 0.002$) (Figure 8).

4. Discussion

4.1. Performance. RTs were statistically significant regarding both load and modality factors, as reported in previous studies (i.e., [94]). Participants' responses were significantly slower during the hardest level (2-back) task than the medium (1-back) and simplest (0-back) levels and during auditory compared to visual tasks (Figures 4(a) and 4(b)). The latter finding conflicts with the hypothesis that auditory stimuli have more durable feature binding [120] and longer lasting representations and thus stimulate enhanced performance [121, 122]. However, there are exceptions to the finding that the auditory condition improves the speed of responses during WM tasks (i.e., [6, 123, 124]). A possible explanation, also advanced by Amon et al. [124], might be that visual stimuli were processed more quickly, but the accuracy scores (90.55% versus 82.33% for auditory and visual conditions, resp.) suggest a more accurate stimulus processing. Furthermore, longer RTs during the auditory condition ($\mu = 731.70 \pm 262.794$) reflected the subjects' perception of difficulty (54.54% of the participants perceived more difficulty with auditory than visual tasks). Another plausible interpretation, complementary to the previous one, could be that visual WM reaches functional maturity earlier than the corresponding auditory system [33]. Response accuracy (ACC) and IES worsened with increasing n-back levels, but the effect of memory load performance was generally more statistically evident in relation to RT than ACC and IES, possibly due to a ceiling effect [125] (Figures 4(c) and 4(d)).

4.2. Electroencephalographic Activation. Neural oscillations provide an effective measure to assess the underlying neural mechanism that enables and controls memory load and memory decay [126]. Previous reports on a quantitative comparison of neurophysiological patterns during different n-back tasks (e.g., [127–131]) involved mostly adult populations (see, e.g., the meta-analyses in [21, 132, 133] study); only rarely did they involve visual and auditory VWM, in particular in children. This study, instead, focused on cortical activation in children during auditory and visual n-back tasks.

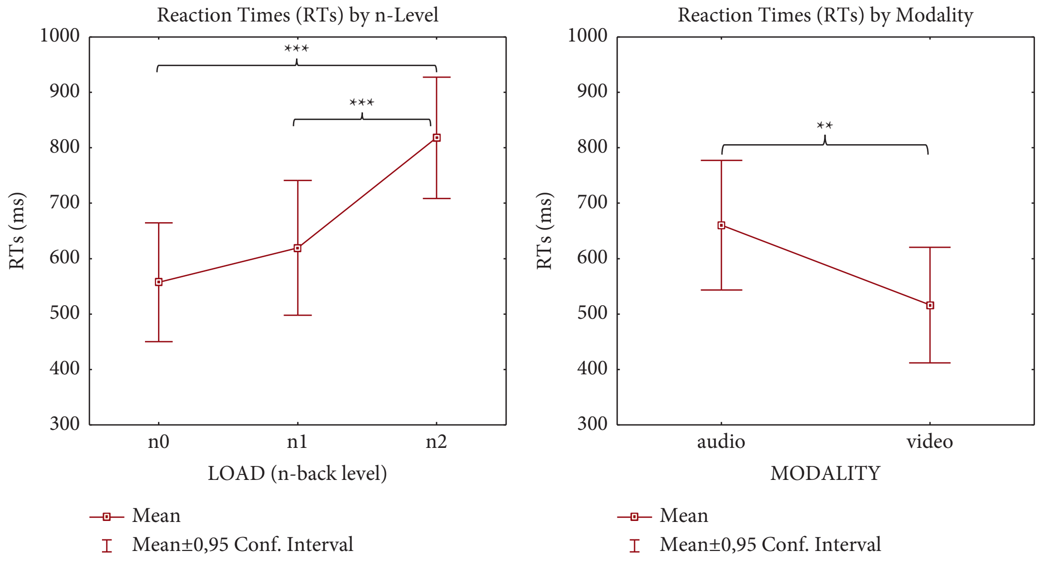
As expected, we found that stimulation of VWM in children appears to activate generally the same brain regions as in adults (Figure 5), albeit in a more widely distributed pattern [38]. Thus, our investigation of EEG differences in stimuli processing during n-back tasks may be an important tool for understanding developing neural functioning.

Table 1: Descriptive statistics of behavioral results.

			MEAN (±d.s.)		
			Load		
		Modality	$n-0$	$n-1$	$n-2$
Behavioral variables	RTs	Audio	646.417 (±231.639)	674.638 (±3.3.793)	874.047 (±202.921)
		Video	468.303 (±228.142)	564.106 (±242.782)	762.179 (±282.728)
	ACC	Audio	0.969 (±0.070)	0.876 (±0.131)	0.870 (±0.108)
		Video	0.909 (±0.166)	0.833 (±0.215)	0.870 (±0.168)
	IES	Audio	664.880 (±218.066)	836.421 (±474.344)	1021.001 (±346.921)
		Video	516.207 (±180.57)	732.827 (±361.078)	1130.646 (±485.263)

Table 2: ANOVA analysis of behavioral results.

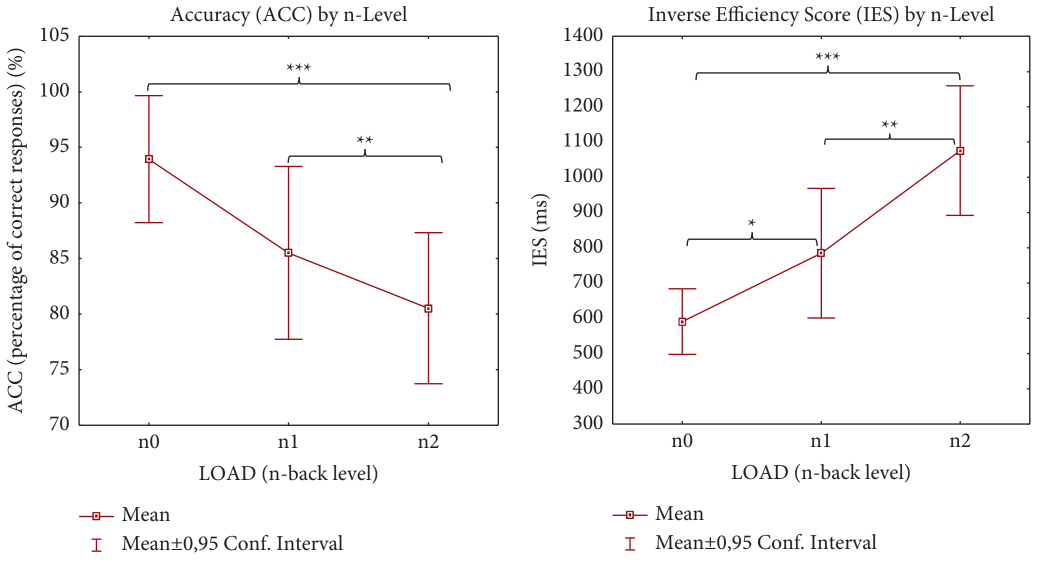
	ANOVA								
	Load			Modality			Load × Modality		
	F	p	η_p^2	F	p	η_p^2	F	p	η_p^2
RTs	25.038	**<0.001**	0.714	10.744	**<0.008**	0.517	1.285	0.298	0.113
ACC	11.7	**<0.001**	0.539	3.036	0.112	0.232	2.189	0.138	0.179
IES	17.671	**<0.001**	0.638	0.371	0.555	0.035	3.421	0.052	0.254



(a)

(b)

Figure 4: Continued.



(c) (d)

Figure 4: Behavioral results. Significantly different ANOVA (see Tables 1 and 2) behavioral results (RT (a) and (b); ACC (c); IES-4 (d)) according to load and modality factors. *Note*. Significant differences between load condition, modality condition, and load x modality condition emerging from the post hoc test are indicated ($^{*}p \leq 0.05$; $^{**}p \leq 0.01$; $^{***}p \leq 0.001$).

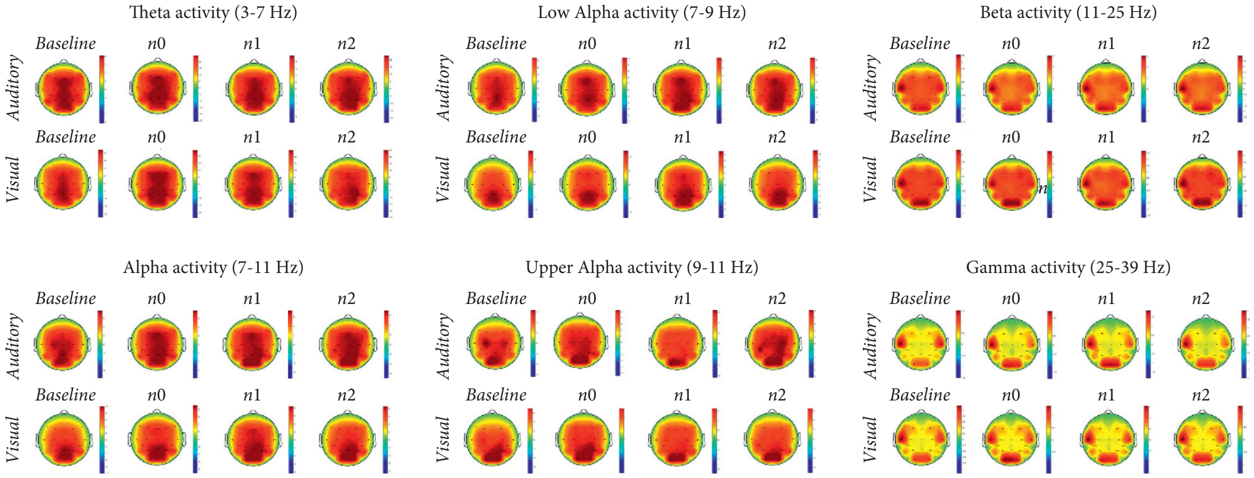
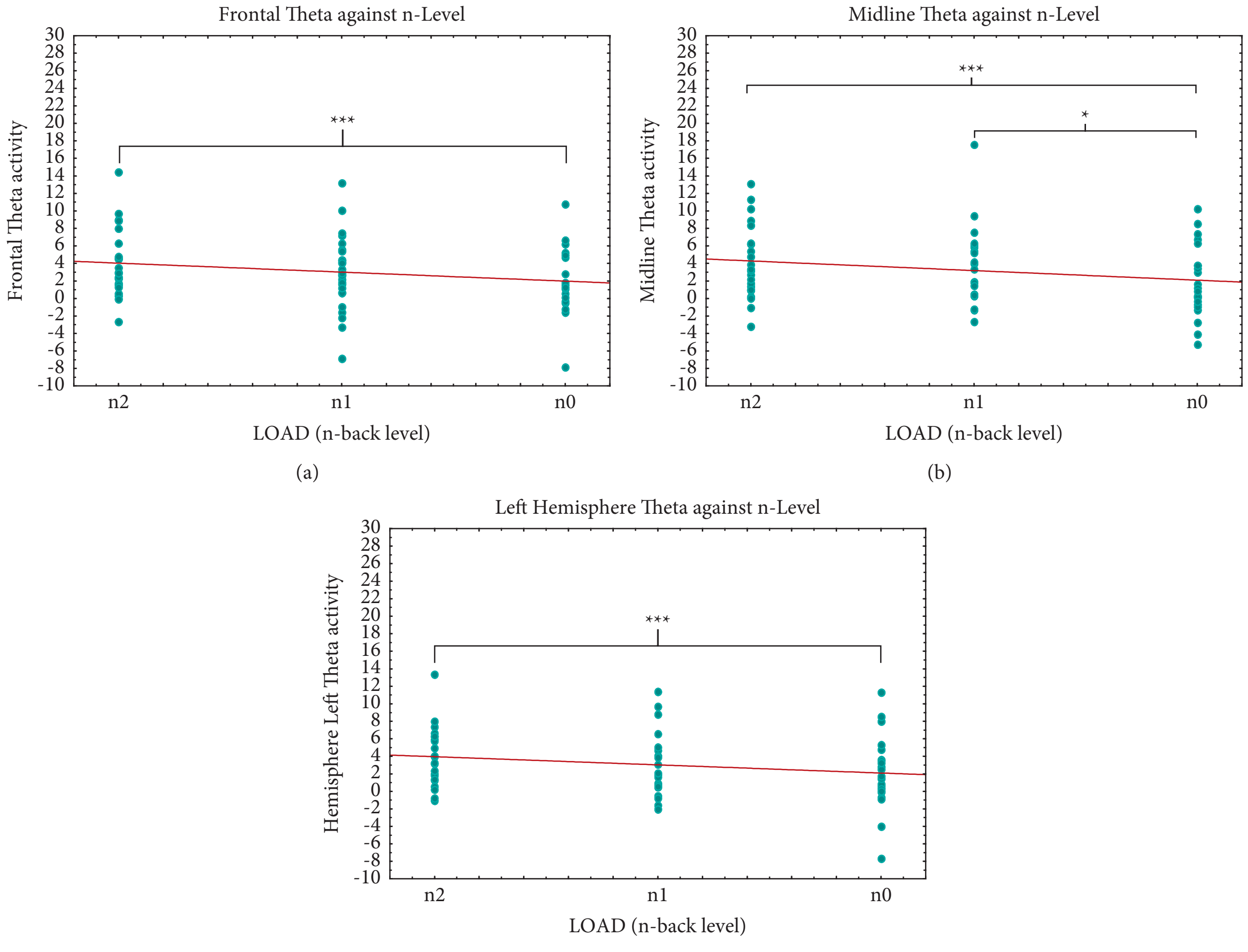


Figure 5: Topographical representation of visual and auditory-verbal working memory in the different frequencies of interest. Topoplots represent the average Power Spectral Density (PSD) of all eleven subjects in the 19 electrodes sites on the scalp for alpha, theta, beta, and gamma frequency bands during the Baseline phase and during each n-back task condition (n-level x modality). Colors describe high (warm-color coded) and low (cold-color coded) intensities of PSD (see color bars).

Table 3: Neurophysiological results of ANOVA analysis.

		Electrode clusters	EEG bands	Load			Modality			Load × Modality		
				F	p	η_p^2	F	p	η_p^2	F	p	η_p^2
Brain areas	Frontal	F3, F4, F7, F8, Fz	Theta	4.6571	**0.021**	0.317	0.672	0.431	0.063	2.273	0.128	0.185
	Midline	Fz, Cz, Pz	Theta	7.697	**0.003**	0.434	1.11	0.316	0.099	2.415	0.114	0.194
	Left Hemisphere	F3, C3, T7, P3, O1	Theta	7.624	**0.003**	0.432	0.016	0.901	0.001	1.958	0.167	0.163
	Parietal	Pz, P3, P7, P8	Gamma	2.021	0.158	0.168	0.018	0.895	0.001	5.851	**0.009**	0.369
	Engagement Index (EI)	Fz, F3, F4, F7, F8, Cz, C3, C4, T7, T8, Pz, P3, P4, P7, P8, Cp5, CP6, O1, O2	Beta/ (Alpha+Theta)	8.674	**0.001**	0.464	0.116	0.74	0.011	0.937	0.408	0.085



(a)

(b)

(c)

Figure 6: Theta band results. Significantly different ANOVA (see Table 3) neurophysiological theta results in different brain areas (resp., in frontal (a); midline (b); and left hemisphere (c) in relation to load. *Note*. Significant differences between load conditions emerging from the post hoc test are indicated ($^{*}p \leq 0.05$; $^{**}p \leq 0.01$; $^{***}p \leq 0.001$).

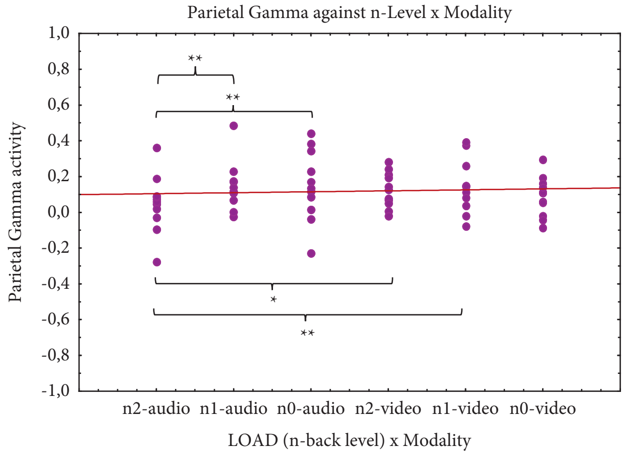


Figure 7: Gamma band results. Significantly different ANOVA (see Table 3) neurophysiological gamma results (parietal brain area) in relation to load and modality. *Note*. Significant differences between load x modality condition emerging from post hoc test are indicated ($^{*}p \leq 0.05$; $^{**}p \leq 0.01$; $^{***}p \leq 0.001$).

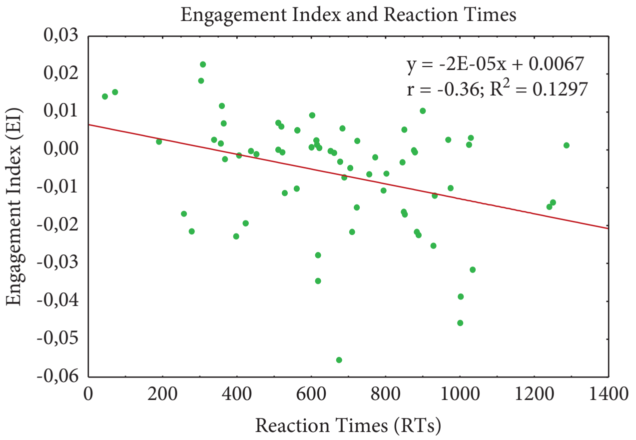


Figure 8: Engagement index and reaction times relationship. Scatter plot showing the negative correlation between engagement index and reaction times values.

We observed a significant increase in theta power in the frontal area related to memory load (Figure 6(a)). This observation is in line with the findings of an EEG study by Gevins et al. [43], reporting the relationship between the increase in frontal theta activity and the task difficulty in subjects performing an n-back task. One interpretation of this activation pattern can be that the increase reflects enhanced attention [45, 134, 135] or effortful cognitive processes [136, 137]. Moreover, multiple function neuroimaging studies have shown that some areas of the PFC are engaged in maintenance and recall of WM representation [14].

EEG evidence of human theta band activity is maximal on the scalp close to the frontal midline; it is often present during waking and is stronger on average during various types of demanding cognitive tasks [43, 138]. Significant theta power changes in the midline area (Figure 6(b)) under various conditions show that this pattern increases with memory load, in agreement with previous studies demonstrating that theta band power in frontal midline scalp increases with mental effort [45, 138–140]. We note that, in our investigation, there was no evidence that theta band activity in frontal and midline areas was influenced by modality (auditory or visual) (Figures 6(a) and 6(b)), results that appear to support our prediction of an a-modal processing of VWM.

The study included exploratory analysis to investigate possible hemispheric lateralization of auditory and visual VWM. It is known that children show hemispheric lateralization in the left frontal and temporal lobes during the VWM task [38] and greater activation of spatial WM in the right frontal, parietal, and occipital cortices [141]. Thus, there may be hemispheric asymmetry for verbal and spatial WM [22], but to date, there have been no reports of investigations on specific modality dissociation of VWM in children. Our results showed a significant strength of lateralization of theta activity in the left hemisphere related to increasing task difficulty but no significant variations related to different task modalities (Figure 6(c)). Lack of influence of audio or video modality on verbal WM is also supported by the absence of significant differences in activation on the F7 electrode that coincides with the Brodmann [142] areas 44–45 corresponding to the Broca [143] language area [144, 145]. The absence of differences in activation of this area related to modality, auditory or visual, could be an indication that VWM is processed as language regardless of stimulus modality in our sample.

Gamma band is another candidate for an EEG signature of WM load [146]. There is evidence that gamma oscillations are involved in perception [147, 148] and are thought to reflect processes related to activation and maintenance of neuronal object representations [149]. Data also suggest a role of gamma in WM as well as perception [150]. Several studies associate this band with higher-cognitive processes [151–154]. Studies have also shown that, in addition to perceptual processing, gamma band activity accompanies many other important cognitive functions such as attention [155–157], arousal [158], language perception [159], and object recognition [160].

Our results show decreasing gamma power in parietal areas to audio stimulation within n-levels (Figure 7). Indeed, the activation trend is inversely proportional to the audio task level, whereas no significant evidence is observed for the video task within n-levels. Comparison of the findings regarding responses to the two different sensory modalities leads us to hypothesize that gamma activity in the parietal cortex is the strongest during the simplest audio condition (0-back) and decreases in the most complex task (2-back). The stronger activation observed during the auditory task (but not visual) seems to be the opposite of the findings of an fMRI study [6]. However, this latter study differed from ours in relation to both the neuroimaging technique used as well as the experimental sample (adults instead of children) and the type of stimuli administered. On the other hand, our finding partially agrees with those of other studies that report enhanced unisensory auditory gamma band activity [161, 162]. Thus, the differences found in parietal areas seem to be of "sensorial origin" rather than strictly connected to the cognitive task. This hypothesis is in line with Karakas et al.'s [163] results showing that the gamma response in the 100 ms after stimulations (in different tasks) has a sensory origin, independent of cognitive tasks. Therefore, in our study, the gamma differences observed in parietal cortices could be attributed directly to the sensory and noncognitive components connected to the VWM task. This result is partially in line with findings supporting consistent variations in gamma activity in relation to memory loads [146] and promotes the hypothesis that parietal regions are part of a network of brain areas that mediate short-term storage and retrieval of phonologically coded verbal material [164]. One might propose that, for both modalities, stimuli appear to be processed in essentially the same regions during the verbal WM task. This idea, as observed by Crottaz-Herbette and colleagues [6], is consistent with the Baddeley model of WM [5, 165, 166], which proposed that both visual and auditory-verbal stimuli are translated into a code stored and manipulated in the phonological loop. This interpretation is further supported by our finding of no significant differences in gamma activation for different modality stimulation in either the Broca area (i.e., language region) or temporal areas (i.e., auditory-verbal regions), which are active during the processing of visual or auditory-verbal representation, respectively [167].

Emotions are omnipresent in human life. Learning is strictly related to emotions [168], and cognitive processes are greatly intertwined with emotional states [169]. Research on emotion and WM has focused primarily on adult clinical populations [170], but some studies investigated emotion-cognition interaction in both clinical and nonclinical populations across development [171, 172]. Other studies have demonstrated that the association between negative affect and academic performance in school is mediated [173] or moderated by WM functioning [174]. Chaouachi and colleagues [175], in order to study the learner's affective changes on the value of EI, found that emotional states are strongly correlated with the learner's EI; therefore, the evaluation of EI may facilitate in-depth investigation of the eventual impact that affective changes have on cognitive processes [175]. Our results showed that EI values decrease for each level of difficulty (from 2 to 0-back). Interestingly, this trend is inversely correlated to behavioral performance data (RTs) (Figure 8). Although the statistical analysis did not reveal a strong correlation between performance and EI, our result is in line with the above-mentioned study by Chaouachi et al. [175] that demonstrates the validity of EI as an indicator of learner performance and suggests the effectiveness of EI also in memory tasks involving children**.** Thus, considering that also the emotional factor is crucial in learning, we can speculate that, in a pedagogical intervention strategy aimed at optimizing the WM process, mental engagement should be taken into account in addition to other behavioral performance indicators. The absence of a statistically significant impact of the sensorial modality on EI could be an important factor in the development of pedagogical intervention aimed at enhancement of cognitive functions even in clinical populations with sensorial deficits.

5. Conclusion

Our findings were consistent with our predictions. Our hypothesis for the identification of an a-modal neural mediation of the theoretical phonological loop which underpins auditory-visual VWM is comprehensively supported. Specifically, the results confirm our double expectations:

(1) Although the same brain areas appear to be involved in both auditory and visual VWM, there were no significant differences in the activation of neural signals with the two modalities, suggesting cross-modal processing of VWM in children.

(2) The strongest significant differences in EEG activation in responses to auditory and visual n-back WM tasks depend on memory load variation. Moreover, the correlation between EI and RT results suggests how the simultaneous study of physiological and behavioral variables related to VWM could be an effective tool to enhance learning in children.

To the best of our knowledge, the present study is the first attempt to identify a neurophysiological benchmark of auditory and visual VWM in healthy children, and the results pave the way to the understanding of fine sensory influences on VWM. However, the present study is not without limitations, like the size of the sample analyzed and the average age of the participants, which means that the results could not be generalized to an older population. Moreover, we are aware that the use of 20 EEG channels cannot allow the precise indication of the areas corresponding to the activation detected at the selected 20 electrodes. Further studies on larger populations and subjects with particular clinical and sensorial conditions could contribute to the identification of eventual specific deficits and to the elaboration of training for target enhancement of WM development in childhood. Finally, the use of an experimental setup with more than 20 EEG channels could offer further developments to these first results.

Data Availability

The raw data supporting the conclusions of this article and the material will be made available by the authors without undue reservation. None of the experiments was preregistered.

Conflicts of Interest

The authors declare that they have no conflicts of interest.

Authors' Contributions

Bianca Maria Serena Inguscio and Giulia Cartocci contributed equally to this work. Fabio Babiloni and Patrizia Mancini equally supervised the project. Bianca Maria Serena Inguscio and Giulia Cartocci contributed to conceiving and conducting the study. Bianca Maria Serena Inguscio prepared the experimental protocol, recorded and elaborated the data, performed the analysis, and wrote the paper. Nicolina Sciaraffa analyzed data. Claudia Nasta and Andrea Giorgi recorded the data. Maria Nicastri and Ilaria Giallini provided support for the organization and realization of the study. Giulia Cartocci and Patrizia Mancini edited the manuscript. Antonio Greco, Patrizia Mancini, and Fabio Babiloni supervised the entire experiment.

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brain sciences

Article

Gamma-Band Modulation in Parietal Area as the Electroencephalographic Signature for Performance in Auditory–Verbal Working Memory: An Exploratory Pilot Study in Hearing and Unilateral Cochlear Implant Children

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Abstract: This pilot study investigates the neurophysiological patterns of visual and auditory verbal working memory (VWM) in unilateral cochlear implant users (UCIs). We compared the task-related electroencephalogram (EEG) power spectral density of 7- to 13-year-old UCIs ($n = 7$) with a hearing control group (HC, $n = 10$) during the execution of a three-level n-back task with auditory and visual verbal (letters) stimuli. Performances improved as memory load decreased regardless of sensory modality (SM) and group factors. Theta EEG activation over the frontal area was proportionally influenced by task level; the left hemisphere (LH) showed greater activation in the gamma band, suggesting lateralization of VWM function regardless of SM. However, HCs showed stronger activation patterns in the LH than UCIs regardless of SM and in the parietal area (PA) during the most challenging audio condition. Linear regressions for gamma activation in the PA suggest the presence of a pattern-supporting auditory VWM only in HCs. Our findings seem to recognize gamma activation in the PA as the signature of effective auditory VWM. These results, although preliminary, highlight this EEG pattern as a possible cause of the variability found in VWM outcomes in deaf children, opening up new possibilities for interdisciplinary research and rehabilitation intervention.

Keywords: working memory; deafness; cochlear implants; children; EEG; gamma; theta; n-back; verbal audio; verbal video

1. Introduction

It is well known that the relationship between hearing and language development is essential in the early years of a child's life (i.e., [1,2]). The perception, development and use of human language are firmly based on an acoustically transmitted signal [3,4]. Therefore, even a slight hearing loss negatively affects language development in children and delays the acquisition of language, social, academic, and sensory skills [5]. Furthermore, as language and speech development are prerequisites for cognitive development, a hearing defect may influence and impair the cognitive abilities of deaf and hard-of-hearing (DHH) children [6].

The benefits of cochlear implantation for restoring hearing and supporting the development of communication skills in prelingually deaf children are firmly established [7–9], especially when using basic clinical measures of speech recognition outcomes. However, there is a tremendous degree of variability and individual differences in the effectiveness of cochlear implants (CIs) [10]. In fact, despite children who receive a CI early in life generally performing better on a wide range of speech and language outcome measures than children who receive a CI at older ages [11,12], significant variability in speech and languages outcomes are routinely observed in this clinical group, and a subgroup of children with CIs fail to attain optimal speech–language outcomes [12,13]. Researching the precise causes of this variability, the extensive studies of the Pisoni and Kronenberger group showed that CI users are at risk of delays in developing Executive Functions (EFs) [8,14–16]. EFs are referred to as higher-order cognitive processes that enable, for instance, to flexibly set up and monitor goal-directed behaviors like attention and regulation, especially in complex circumstances [17,18]. Children with CIs showed greater rates of delay than hearing peers in multiple subdomains of EFs. However, it is in verbal working memory (VWM)—commonly defined [19] as the temporary maintenance of verbal information (i.e., some aspects of language)—that the most significant and most consistent delay, compared to hearing peers, has been found [8,20–24], regardless of the modality of the verbal stimuli presentation [25]. As a result, VWM has been identified as a fundamental domain of neurocognitive risk and a potential target for intervention to enhance speech–language outcomes in CI users [24]. However, a comprehensive understanding of how these factors interact and the neural mechanisms underlying these interactions are unknown. In fact, most studies have addressed working memory (WM) deficits in children with cochlear implants by assessing outcomes with psychometric scales and speech tests [26,27], especially in Verbal WM, and investigation of the neural correlates of this crucial executive function in children is rare, particularly in clinical children's samples [28].

Early sensory experiences shape the neural circuitry of the auditory system, and there is a pronounced reduction in synaptic plasticity in the auditory cortex during early deafness [29]. Electrophysiological studies have shown that early access to sound with CI can mitigate some structural and functional effects of congenital deafness on the cortical auditory system [30], which are likely to change the cortical network involved in hearing [31,32]. However, CIs provide poor sound encoding in terms of frequency selectivity and temporal encoding if compared to hearing subjects [33]. These interventions are thus are unlikely to completely restore the normal auditory connectome, even in children who receive implants in both ears at young ages. So, as Kral and Sharma [34] pointed out, a more comprehensive understanding of the neural correlates of individual variability will be critical to developing better rehabilitation options that are aimed at and customized for individual patients. Moreover, neuroimaging studies have broadly observed cross-modal plasticity in DHH subjects with CI [35,36]. For example, Song and colleagues, in a positron emission tomography (PET) study [37], found greater visual activation for audiovisual speech in CI users, suggesting a plastic effect on multimodal perception, inferring an incomplete reversal of cross-modal plasticity after hearing restoration, or that auditory reorganization is maintained by a continued reliance on visual input [38]. In a functional magnetic resonance imaging (fMRI) study with early deaf adults, auditory regions showed greater cross-modal activation during a visuospatial WM task in clinical subjects than in controls. Furthermore, cross-modal activation in the auditory areas correlated with WM performance in deaf but not in hearing participants [39]. Considering these findings and the previous literature on left-hemispheric lateralization of WM in hearing subjects [40,41], the question arises as to the lateralization of VWM function and whether or not it is dependent on the sensory modality of stimulation in DHH children.

Few recent studies have investigated cortical activations through electroencephalography (EEG), a powerful, accessible and versatile neuroimaging tool for investigating human brain physiology, cognition and behavior [42,43] in CIs children during cognitive tasks. For example, researchers used theta and alpha connectivity to differentiate performance across different implant processors [44] or hemispheric gamma activation to suggest the occurrence of a sensitive period for CI surgery for best emotion recognition skills development [45]. Moreover, during a listening task in the noise, a higher workload index (theta/alpha) in over-demanding situations is recorded [46], as well as displaying lower parietal alpha power levels in the most challenging listening condition [47]. Furthermore, alpha oscillations appear particularly sensitive to hearing loss during WM paradigms [48]. Finally, Cartocci and colleagues [49] showed a correlation between the period of deafness and the cortical activity asymmetry toward the hearing ear side in the frontal, parietal and occipital areas. These findings are in accord with several studies reporting that strain on cognitive resources for auditory perception leaves less availability for cognitive processing [50–52] and converge toward the hypothesis that the cause of the wide variability found in deaf children with cochlear implants in VWM may lie in scalp-recorded neural oscillations. Indeed, there has been considerable interest in determining the locus of outcome variability in children with CI in recent years. Researchers have identified that cognitive factors, such as WM, are critical in the healthy development of children with CI, and despite the effect of CI-treated profound hearing loss on auditory and visual–verbal WM performance, its neural correlates remain unclear [48,53]. In particular, to our knowledge, the EEG power spectrum related to the audio-visual WM task has not been explored in children CI users.

The Aim

In light of the scientific evidence available in the literature to date, the present study aims to investigate the impact of unilateral cochlear implant use in deaf and hard-of-hearing (DHH) children on the neurophysiological patterns underlying VWM processing of auditory and visual stimuli during an n-back task. The pioneering goal of the present study, which is comparing the EEG signals of DHH children unilateral CI users with a control group of hearing children, is to reveal the neurophysiological sensorial patterns of VWM. This is in order to explain the extreme behavioral variability found in this clinical group concerning this essential cognitive function. Findings could support an improved clinical and rehabilitative approach for these patients.

2. Materials and Methods

Methods for recording and analysis in the current study follow those described in previous publications from our group [28,45]. An abbreviated version of these methods is provided below.

2.1. Participants and Ethics Statement

The sample size was determined by a power analysis before data collection using G*Power (Universität Düsseldorf, Düsseldorf, Germany) [54]. Given the preliminary nature of the present study, seven right-unilateral cochlear-implanted children (UCI, mean age 11.22 years ± 0.63 SD) and ten age-matched hearing children (HC) were recruited. Still, this went beyond the minimum 10% of the total sample requested for pilot studies [55]. Demographic and clinical data for the UCI group are summarized in Table 1. The eligibility criteria for the clinical group included congenital severe/profound deafness (Pure Tone Average in the better ear $\geq$ 80 dB HL for 500–4000 Hz), good speech perception abilities, defined as bisyllabic word recognition and sentence comprehension >90% in a silent room at the moment of the EEG test; none of UCI wore any hearing aid in the contralateral ear to the one with the cochlear implant. The age of the sample was determined according to previous studies [56,57]. Raven's standard progressive matrices (RPM) [58], a test of non-verbal spatial reasoning, was used for the screening for the participant selection. Exclusion criteria for enrolment in the study were diagnosis of neuropsychiatric disorders and/or sensory deficits; children with scores below the standard average for their age (taken from test norm) on RPM; left-handed children due to past evidence of handedness influence on cerebral laterality [59]. Before the experiment, participants and their parents were fully informed about the study. The investigation was conducted according to the principles outlined in the Helsinki Declaration of 1975, revised in 2000 and approved by the Institutional Ethics Committee of Policlinico Umberto I—Rome, Italy (no. 259/2020). Informed written consent was obtained from all parents before the protocol started. Participation in the study was voluntary; children received a present after their involvement.

Table 1. Demographic and clinical data concerning the unilateral cochlear implant (UCI) group. In particular: onset of deafness, its etiology, and auditory age (years of cochlear implant use since implantation).

Participants	Gender	Onset of Deafness	Degree	Aetiology	Current Age	Age at CI	Auditory Age
P1	F	Congenital	Profound	Homozygous mutation of the connexin-26 gene	12.00	2.90	9.09
P2	F	Congenital	Profound	Homozygous mutation of the connexin-26 gene	10.73	1.86	8.87
P3	F	Congenital	Profound	Homozygous mutation of the connexin-26 gene	11.49	1.41	10.07
P4	F	Congenital	Profound	Homozygous mutation of the connexin-26 gene	11.49	1.41	10.07
P5	F	Congenital	Profound	Homozygous mutation of the connexin-26 gene	11.14	1.16	9.97
P6	M	Congenital	Profound	Usher syndrome	11.58	0.79	10.78
P7	F	Congenital	Profound	Unknown	10.09	1.79	8.30

2.2. Overview of Experimental Design and Procedure

During the EEG recording, participants performed two verbal n-back tasks [60] with different memory loads from 0-back to 2-back: an auditory n-back task (AUD-task) in which stimuli were presented aurally, and a visual n-back task (VIS-task) in which stimuli were presented visually. The order of the task administration and the order of the n-back blocks presentation were randomized across participants.

Stimuli: verbal material consisted of auditory and visual stimuli, referring to seven consonants (c, g, k, p, q, t, v) already used in previous experimental protocols [61–64]. Vowels were excluded to reduce the likeliness of participants developing chunking strategies [65]. To ensure correct perception by UCI and HC groups, we performed a stimuli exposure pretest. Visual stimuli (Consolas font—130) with a duration of 500 ms and an interstimulus interval of ISI 3000 ms [56] were presented one at a time on a grey background in the center of a monitor screen placed at eye level, 50 cm from the participant. Auditory stimuli (duration 500 ms; ISI 2500) [61] consisted of a recorded female voice, set at a 65 dB SPL intensity to ensure comfortable audibility to both HC and UCI [45], transmitted by two audio speakers placed at 45 degrees left/right, at face level 1 m in front of the participant.

Task execution: Immediately after the stimuli presentation, participants in the ISI had to respond by pressing a previously reported key (D/K) on the keyboard to indicate whether the letter was a target (K) or a nontarget (D): thus, there was a behavioral response in either case. In the 0-back condition, the letter X was the target. In the 1-back condition, a letter was a target when it was the same as the one presented immediately before. In the 2-back condition, a letter was a target when it was the same as the two letters before. Participants received detailed instructions on how to perform the task correctly and a training session was undertaken before the practical measurement session to familiarize them with the experimental procedure.

Task structure: load levels (0, 1, 2-back) were presented in six blocks (2 for each level) for each task (auditory and visual). The blocks consisted of 21 randomized stimuli (30% target) [56]. A baseline phase, during which participants were asked to remain relaxed with no task except to look at the screen while auditory or visual stimuli were presented, anticipated the task phase. During the baseline phase, the 7 stimuli were repeated randomly 3 times (500 ms with 3000 ms ISI), creating a 21-item block analogous to the experimental blocks. The task phase then consisted of 2 randomized presentations of the three blocks. Thus, each session consisted of 3-n back levels per 2 presentations for 6 blocks in randomized order for audio and visual tasks. Half of the participants started with the visual stimuli task, and the other half with the auditory task (see Figure 1 for a visual task structure synthesis). A Lenovo PC (monitor resolution 1024 × 768) displayed and controlled stimuli presentation and collected participants' responses in terms of reaction times (RTs) and correct responses (CRs) through the software package E-Prime (Psychology Software Tools, Pittsburgh, PA, USA, Version 3.0).

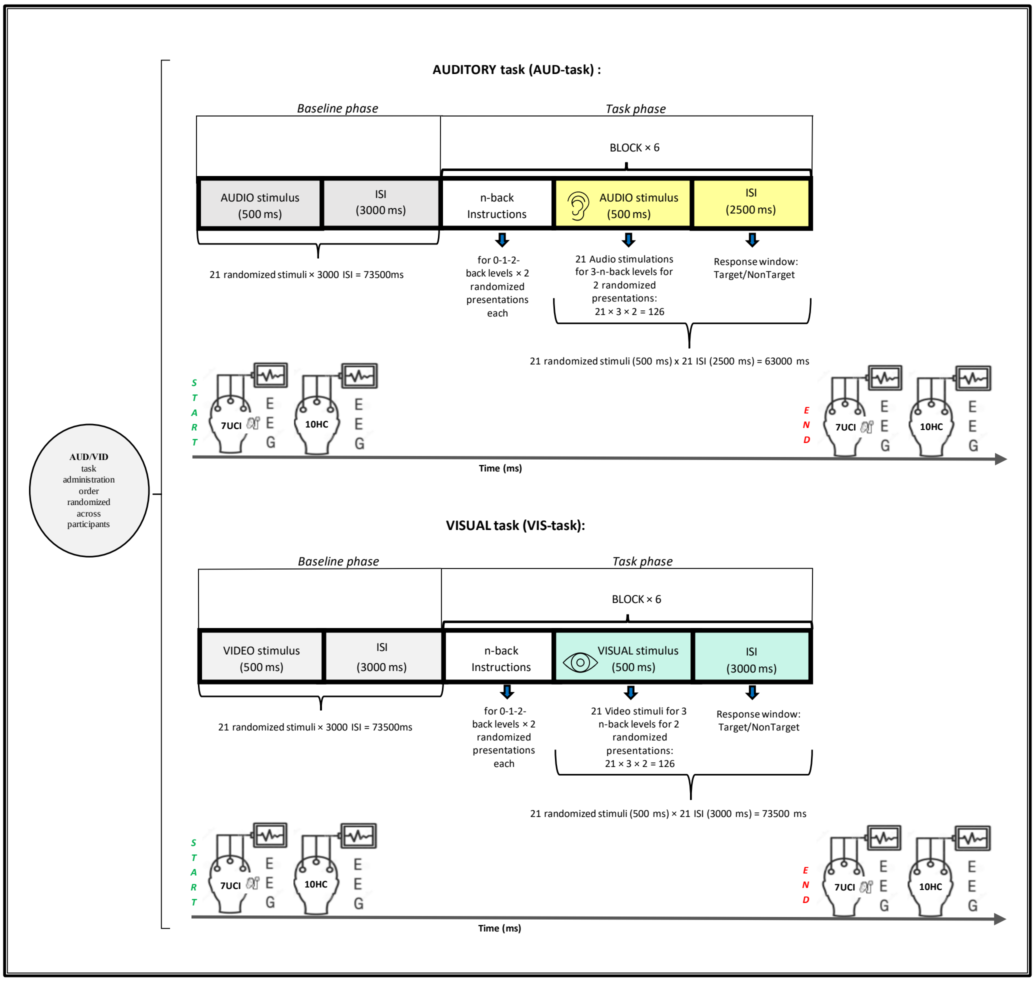

Figure 1. Experimental design with the trial timeline. Schematic illustration for each n-back task (auditory-AUD and visual-VIS modalities) performed by hearing children (HC) and unilateral cochlear implant (UCI) groups during electroencephalography (EEG) recording. Each modality task started with the baseline phase, followed by the task phase.

Procedure: the participant was seated on a chair in an audiometric test room while the experimental design was fully explained. Participants were instructed to assume a comfortable position and avoid unnecessary movement to reduce muscular artefacts in the EEG signal. After each task phase, the participant indicated the perceived task difficulty (easy–medium–hard) on a stylized image; at the end of the entire experimental session, they were asked to rate which of the two tasks (visual or auditory) was the most difficult.

2.3. Behavioral Measures

Performances were evaluated in terms of accuracy (ACC), calculated as the percentage of correct responses for each task condition (each n-back level for auditory and visual modality tasks). To integrate the correct answers and the reaction times (RTs, measured from the time of stimulus offset) for each response, inverse efficiency score IES = RT/1-PE was calculated, where RT is the subject's average RTs for correct answers (target/nontarget). PE is the subject's proportion of errors for each condition. IES can be interpreted as the RT corrected for the number of errors committed [66].

2.4. EEG Recording and Signal Processing

To record 20 EEG channels (Fpz, Fz, F3, F4, F7, F8, Cz, C3, C4, T7, T8, Pz, P3, P4, P7, P8, Cp5, Cp6, O1 and O2) referred to the participants' earlobes, a digital ambulatory monitoring system (BePlus System-EBNeuro, S.p.A., Firenze, Italy) with a sampling frequency of 256 Hz was used. The impedance was kept below 10 kΩ, and a 50 Hz notch filter was then applied to remove power interference. EEG signals were initially band-pass filtered with a 5th order Butterworth band-pas filter (1–45 Hz) to reject continuous components and high-frequency interferences like such as muscular artefacts. The Fpz channel was used to eliminate eye-blink contributions by the REBLINCA algorithm [67] without losing data. Specific procedures of the EEGLAB toolbox (Schwartz Foundation, Halesite, NY, USA) [68] were used to depurate from other artefacts. The EEG dataset was segmented into epochs starting 500 ms before stimulus onset and ending 2500 ms after the offset. This temporal windowing was adopted to respect stationary EEG and allow for a high number of observations, compared to the number of variables considered in the analysis [69]. To identify artefacts, three criteria were employed according to published procedures [45,49,70]: (i) threshold criterion (±80 μV); (ii) trend estimation criterion (slope higher than 40 μV/s or less than 0.3 μV/s); (iii) sample-to-sample criterion (when, in terms of absolute amplitude, the signal sample-to sample >30 μV/s). Finally, all epochs marked as "artefacts" were removed from the EEG dataset, such that all analyses were based on clean EEG signals. To accurately define EEG bands of interest, individual alpha frequency (IAF), given in Hertz, was computed for each participant on a 60 s long-closed eyes segment, recorded before the baseline phase [71]. Each band was then defined as IAF± x, where x was an integer in the frequency domain; thus, the EEG signal was filtered in the following frequency bands in Hertz (Hz): theta $[\mathrm{IAF} - 6 \div \mathrm{IAF} - 2\,\mathrm{Hz}]$, alpha $(\mathrm{IAF} - 2 \div \mathrm{IAF} + 2\,\mathrm{Hz})$, beta $(\mathrm{IAF} + 2 \div \mathrm{IAF} + 16\,\mathrm{Hz})$, and gamma $(\mathrm{IAF} + 16 \div \mathrm{IAF} + 30\,\mathrm{Hz})$ [71]. Then, the power spectral density (PSD) [72] was calculated for each epoch and channel, with a Hanning window of 1 s and an overlap of 500 ms. Topographical distribution of band modulation analysis was based established on averages of the data for the following areas of interest (AOIs): frontal, parietal, occipital and hemispheres electrode locations. The channels considered were F3, F4, Fz (frontal); Pz, P3, P4, P7, P8 (parietal); O1, O2 (occipital); F3, C3, T7, P3, O1 (left hemisphere); F4, C4, T4, P4, O2 (right hemisphere). In addition, the Workload Index (WI) was calculated in accordance with the formula given above.To limit bias on scores due to subjective stimuli perception on VWM n-back task recording, PSD data were normalized with respect to the baseline [73].

2.5. Statistical Analysis

Both neuro and behavioral data were objects of statistical analysis in this study. The Shapiro–Wilk normality test [74] was applied to the dataset under investigation. Then, depending on the results, parametric or non-parametric analysis of variance (ANOVA) was performed [75]. Both behavioral (ACC; IES) and neurophysiological (AOIs; WI) values were entered in a 2 × 3 factorial ANOVA with two factors: factor modality (with 2 levels: audio and video) and factor load (with 3 levels: 0-1-2). Duncan's post hoc test [76] was used to investigate statistically significant results of ANOVA tests; partial eta squared (η_p^2) effect sizes were reported [77,78]. The ANOVA test has sufficient statistical power to deal with the analysis of relatively small numbers of participants, as in this study [79], provided that the number of factors is lower than 4, as in this case. A correlation analysis was performed to assess possible relationships between variables, while simple regression analysis was used to investigate potential functional relationships between variables (e.g., mean of tot audio; mean of total 2 back). A cut-off of $\alpha = 0.05$ was set as the cut-off of significance [80].

3. Results

3.1. Behavioral Results

The overall ACC percentages were greater during all auditory n-back levels and increased with decreasing memory load for both HC and UCI. Furthermore, IES scores for both groups were higher during all levels of auditory n-back and decreased with decreasing memory load compared to visual n-back (Table 2).

Table 2. n-back task behavioral performances of both investigated groups (unilateral cochlear implant users—UCI; hearing control—HC) and the total participants (TOT) in terms of accuracy (ACC) and inverse efficiency score (IES) expressed in milliseconds (ms) for all the experimental conditions.

ACC (%) and IES (ms) for n-Back Task Conditions								
		👂	ACC Audio 0-back	ACC Audio 1-back	ACC Audio 2-back	IES Audio 0-back	IES Audio 1-back	IES Audio 2-back
Groups	UCI		97.62%	82.65%	75.17%	559.83	814.71	1140.49
	HC		97.67%	86.67%	86.20%	660.45	830.72	1018.99
	TOT		97.06%	85.01%	81.66%	619.02	824.13	1069.02
		👁	ACC Video 0-back	ACC Video 1-back	ACC Video 2-back	IES Video 0-back	IES Video 1-back	IES Video 2-back
Groups	UCI		94.90%	81.29%	80.61%	447.78	875.87	894.68
	HC		90.00%	81.67%	74.52%	505.95	738.62	1079.56
	TOT		92.02%	81.51%	77.03%	482.00	795.13	1003.43

ANOVA results in terms of ACC showed a statistically significant difference between memory load (F(2,30) = 23.992, $p < 0.001$, $\eta_p^2 = 0.615$). The highest significant percentages of correct responses were measured for n0 compared to n1 ($p < 0.001$) and n2 ($p < 0.001$) load conditions (Figure 2).

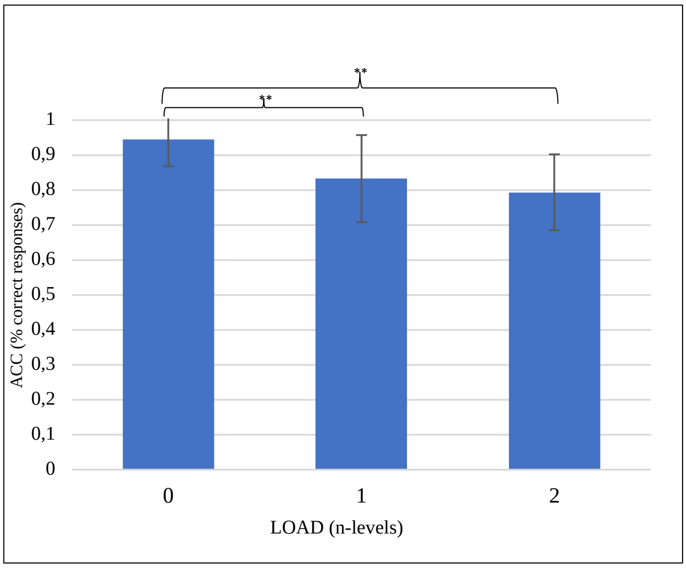

Figure 2. The graph shows the significantly different ANOVA behavioral results of performances in terms of accuracy (ACC) expressed as % of correct responses according to LOAD condition (the 3 levels of the n-back verbal working memory task). Significant differences between load conditions emerging from the post hoc test are indicated (** $p \leq 0.01$).

ANOVA results showed a statistically significant effect of both LOAD (F (2,30) = 30.351, $p = 0 < 001$, $\eta_p^2 = 0.669$) and MODALITY (F (1,15) = 5.8843, $p = 0.028$, $\eta_p^2 = 0.281$) and a significant interaction between these two factors (F (2,30) = 7.3515, $p = 0.002$, $\eta_p^2 = 0.328$) on IES. Post hoc analyses revealed significant increases in IES, respectively, both as the difficulty increased: from level 0-back to level 1 and 2-back ($p < 0.001$ resp.) and between level 1 and 2-back ($p = 0.024$) and for the auditory compared to visual modality ($p = 0.028$) (Figure 3). Furthermore, the 2-audio condition produces significantly higher IES values than all task conditions ($p < 0.001$).

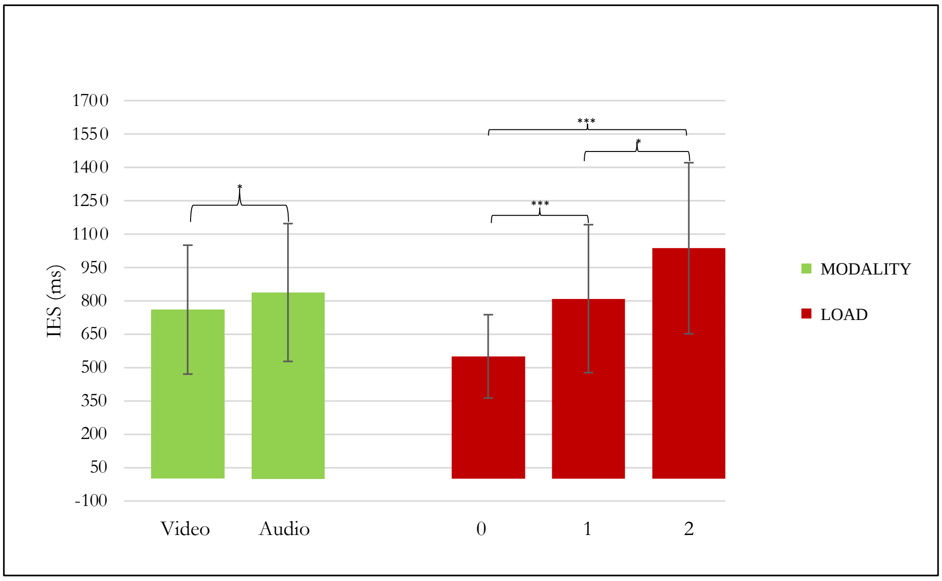

Figure 3. The graph shows the significantly different ANOVA behavioral results of inverse efficiency score (IES) in milliseconds (ms) according to LOAD (the 3 levels of the n-back verbal working memory task) and MODALITY (video and audio) conditions. Significant differences between verbal working memory (VWM) load and modality conditions emerging from the post hoc test are indicated (* $p \leq 0.05$; *** $p \leq 0.001$).

3.2. Neurophysiological Results

When the EEG theta band was considered, there was a significant effect of the LOAD condition on power values (F (2,30) = 4.852, $p = 0.014$, $\eta_p^2 = 0.244$) in the frontal area. In this case, post hoc Duncan's test detected a significant difference between 2-back and 0-back levels ($p = 0.002$) (Figure 4).

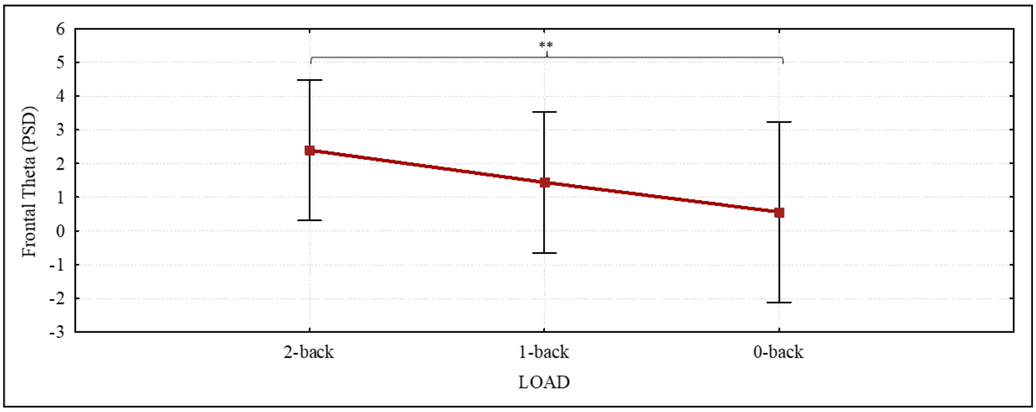

Figure 4. Theta band results. The graph shows the significantly different ANOVA power spectral density (PSD) theta results in the frontal area in relation to verbal working memory (VWM) n-back task levels (LOAD). Significant differences emerging from the post hoc test are indicated (** $p \leq 0.01$).

Concerning gamma activity, the analyses in the parietal area showed considerable interaction between LOAD × MODALITY × GROUP factors (F (2,30) = 3.499, $p = 0.043$ $\eta_p^2 = 0.189$). Post hoc analyses showed a significant difference solely within HC between condition 2 audio and all other conditions ($p \leq 0.001$) except for the 0-video condition (Figure 5).

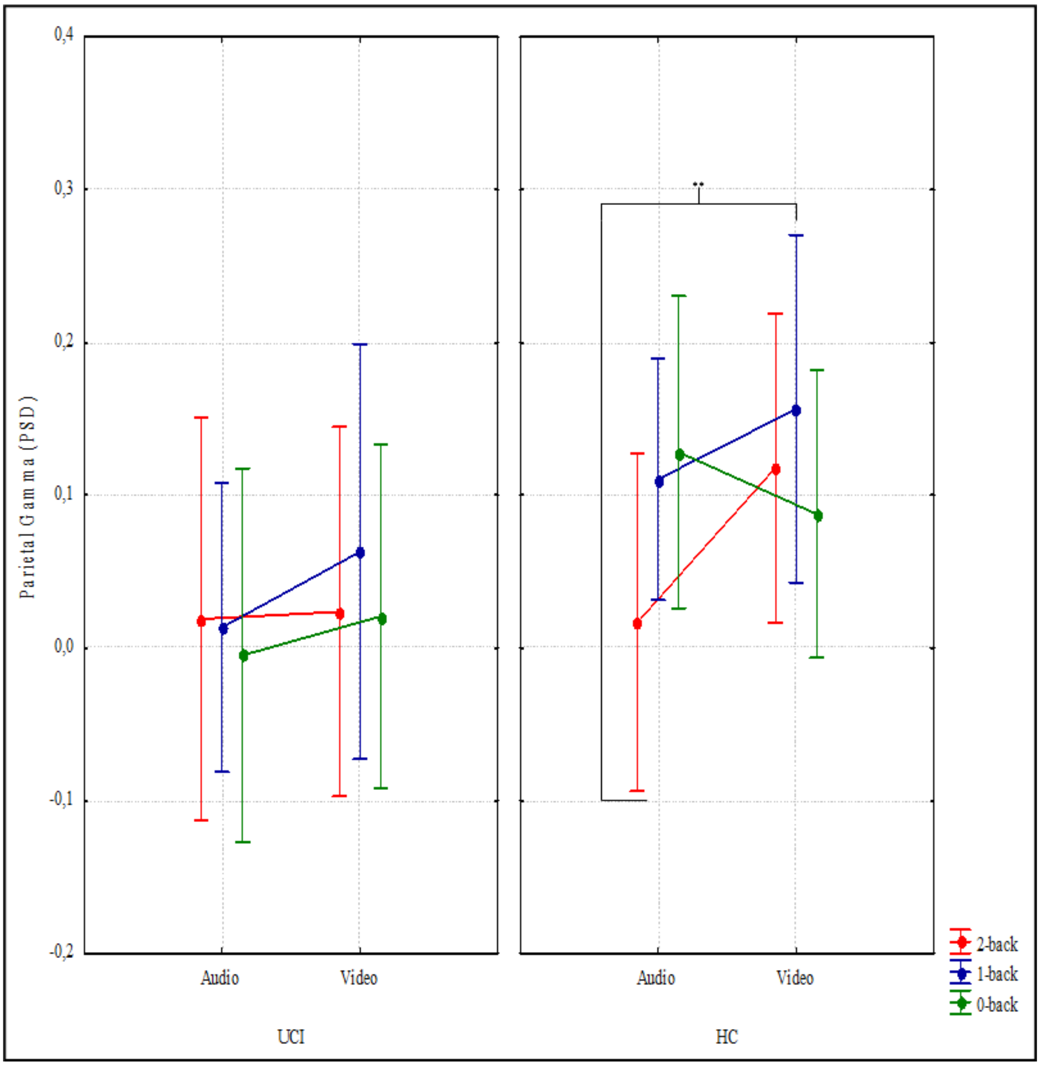

Figure 5. Gamma band results. The graph shows the significantly different ANOVA power spectral density (PSD) gamma results in the parietal area in relation to LOAD (n-back task levels) × MODALITY (Audio and Video) × GROUP (HC = hearing children; UCI = unilateral cochlear implanted children). Significant differences between conditions emerging from the post hoc test are indicated (** $p \leq 0.01$).

Gamma activity also, regardless of conditions, was greater at the limit of significance in the left hemisphere than in the right (F (1,15) = 4.165 $p = 0.059$, $\eta_p^2 = 0.270$). At the same time, this activation pattern in other frequency bands was not observed. Delving into the different hemispheric activation between groups based on bands, solely for gamma, a marginally significant difference is observed (F (1,15) = 4.4381, $p = 0.052$, $\eta_p^2 = 0.228$), revealing a significantly greater activation in the left hemisphere for HC compared to UCI (see Figure 6 for the global activations in gamma frequency).

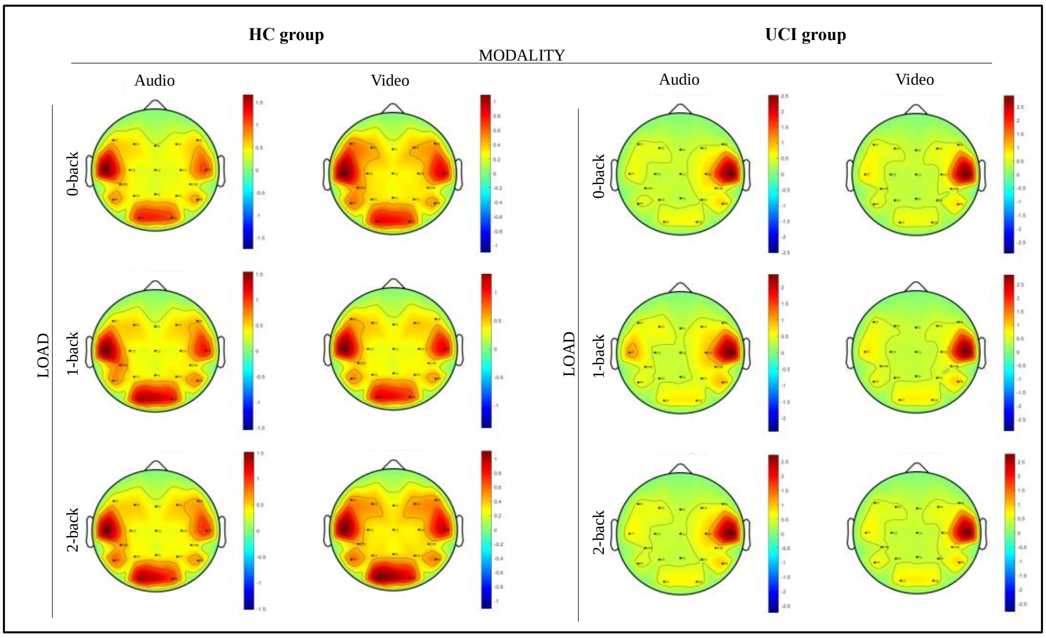

Figure 6. Scalp distribution of the Electroencephalographic (EEG) gamma spectral power during the audio and video n-back tasks. Taped to the left is the gamma activation of the hearing control (HC) group, and to the right, those of the unilateral cochlear implant (UCI) group. For each group from left to right, the scalp maps correspond to the MODALITY (audio and video) and LOAD (0, 1, 2 level) n-back verbal working memory (VWM) task conditions. The black dots correspond to the electrode positions.

In terms of WI, the analysis showed the effect of both LOAD (F (1,15) = 8.679 $p = 0.010$, $\eta_p^2 = 0.206$) and MODALITY (F (1,15) = 8.679 $p = 0.010$, $\eta_p^2 = 0.366$) factors. Post hoc analysis highlighted increased WI values comparing 2-back and 0-back levels ($p = 0.002$) and during video versus audio presentation ($p = 0.008$) (Figure 7).

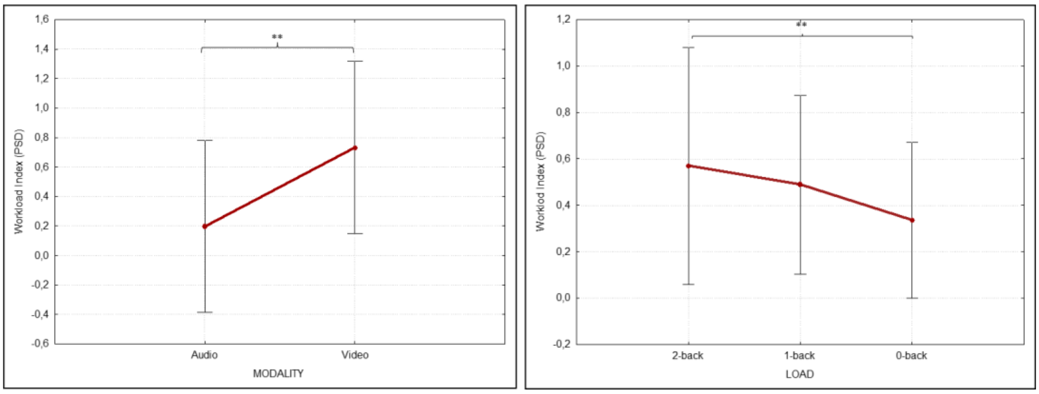

Figure 7. Workload Index (WI) results. Significantly different ANOVA power spectral density (PSD) WI results in relation to both MODALITY (**left**) and LOAD (**right**) conditions. Significant differences between conditions emerging from the post hoc test are indicated (** $p \leq 0.01$).

Correlation analysis showed significant relationships between parietal activation for audio conditions in gamma band and audio IES in HC (r = −0.71), while in UCI, a correlation is observed only between parietal gamma activation with current age for audio condition and not with behavioral data (r = 0.86). Furthermore, during the audio task, always considering the limitations of the analysis due to the numerosity and variability of the group, the simple linear regression analysis demonstrated a significant linear dependence between gamma oscillations in the parietal area and IES only for HC participants ($R^2 = 0.506$, R^2 adjusted = 0.445) and during the audio task (Figure 8).

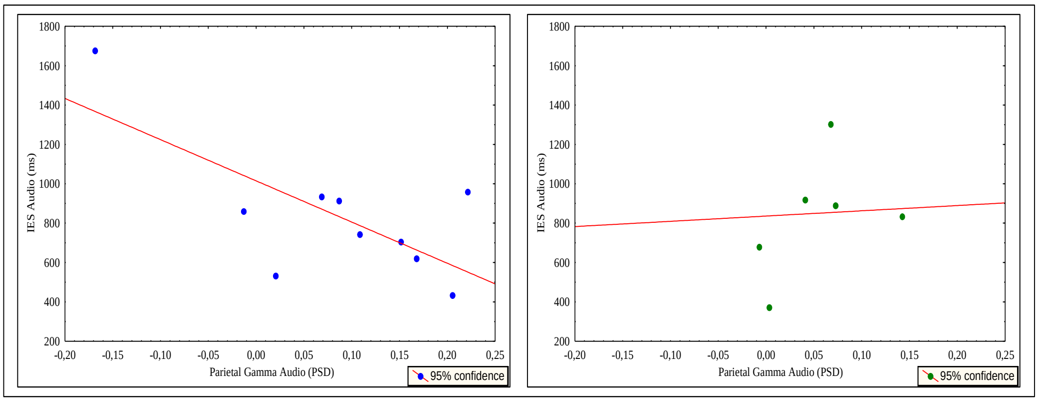

Figure 8. Scatterplot of auditory inverse efficiency score (IES) as predicted by parietal gamma activation across the hearing control (HC) group (*n* = 10) (left, blue dots) and not predicted across the unilateral cochlear implanted (UCI) group (*n* = 7) (right, green dots). Simple linear regression explained 50.6 % of the variance in IES performance based on parietal gamma activity during the auditory n-back task for the HC group.

Inversely, UCI participants showed a linear dependence between current age and EEG activation in the gamma band during the audio task ($R^2 = 0.741$, R^2 adjusted = 0.689) (Figure 9).

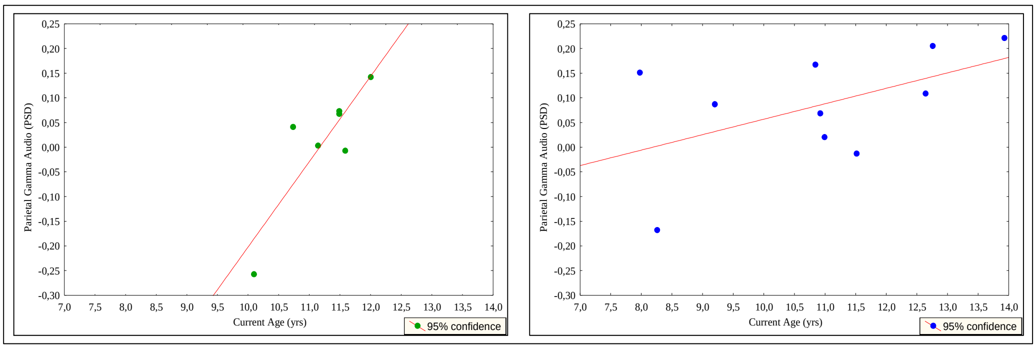

Figure 9. Scatterplot of parietal gamma activation as predicted by current age across the unilateral cochlear implant (UCI) group (*n* = 7) (left, green dots) and not predicted across the hearing control (HC) group (right, blue dots). Simple linear regression explained 74.10% of the variance in parietal gamma activation based on the current age auditory n-back task for the UCI group.

4. Discussion

In the current pilot study, we analyzed behavioral and neural correlates of VWM processing during auditory and visual n-back tasks in normal hearing and DHH children unilateral cochlear implant users. To date, these measures have rarely been used in UCI children while being reported for the visual n-back task in hearing adult and children's groups (i.e., [57,64,81,82]). Primarily, this study is the first one in which the same participants (HC and DHH) conducted a verbal n-back task in two sensory modalities (auditory and visual) while EEG data were recorded. This allowed us to compare the typical load-related EEG (i.e., alpha, theta, gamma) and behavioral measures for two different tasks and between normal hearing and deaf groups. Overall, we expected these measures to show significant differences between groups to explain the extreme VWM behavioral variability found in children receiving cochlear implant.

4.1. Behavioral Results

Predictably, performance in terms of accuracy (ACC) worsens as the VWM load increases (Figure 2) in line with the general literature on n-back tasks children's performances [56,63]. Furthermore, the sensory modality does not influence performances in both groups. In fact, in contrast to the prior research that consistently demonstrated poorer VWM skills in children CI users [24,83], no effect of group membership is observed on both accuracy and IES measures. However, it is essential to note that VWM in the studies mentioned above was individually assessed by batteries of psychometric tests (i.e., subtest of WISC-V). Whereas, in the present study, we used an n-back task, a handy tool for the investigation of the WM process, especially in children [56]. Therefore, since the same assessment instruments were not used, the direct comparison of the behavioral data cannot have absolute reliability. Moreover, both HC and UCI groups have higher IES values in the audio conditions, especially for 2-back, which seems to be the most challenging task condition [84] regardless of the group (Figure 3). Longer IES values for the auditory task are in line with previous studies on HC children and adults [28,85–87], and contradict the hypothesis that auditory stimuli enhance performance by having longer-lasting representation [88,89] and more durable stimuli binding [90]. This result seems particularly interesting regarding the clinical group, given that the model of Pisoni and colleagues [23] proposes that poor performance on VWM tasks in CI users could be partly due to fragile, underspecified phonological representations of letters in short-term memory. Globally, behavioral VWM performances were not different between CI users and hearing controls. However, evidence suggests that performance differences between VWM tasks in post-lingually deafened CI users (both adults and children) and HC are mixed [91,92]. For example, our results are consistent with those reported by [48] during a visual WM task in adults. Furthermore, the latter involved only visual stimuli and concerned an adult sample. Therefore, the lack of differences in performance with DHH children and the absence of significant correlation with the auditory age would preliminarily confirm the cochlear implant's effectiveness in supporting deaf children's VWM performance during an n- back task, regardless of the modality of stimulation.

4.2. Neurophysiological Results

4.2.1. Workload Index

Mental workload (WL) is a fundamental concept in the study of human performance, emerging from the observation that our cognitive system has a limited capacity to perform a cognitive task [93,94]. WL emerges from the interaction between the task at hand and the individual with limited resources [95]. A previous study reported a WL index (WI) modulation in CIs children during the most challenging noise condition in a forced-choice word recognition task [46]. It is, therefore, to be expected that CI users would show a higher WI in the auditory than in the visual n-back task in light of their clinical condition.

To infer the mental state of cognitive load in VWM, we combined the neurophysiological WI [71,96] with a self-report measure of subjective difficulty. During the n-back task, the WI estimate reflected in part the trend of the behavioral IES score. In fact, WI presented the highest values during the 2-back and the lowest values during the 0-back, respectively, the most and least challenging WM conditions, with no differences between the HC and UCI groups (Figure 7). Furthermore, the increased WI in the video condition compared to the audio condition (Figure 7) reflected the total perception of self-reported difficulty by 52.94% of the global participants. Moreover, while self-reported difficulty was equally distributed between auditory and visual modalities in the HC group, 57.142% of the UCIs stated that they perceived more difficulty with the visual task than for the auditory task. Our findings, although preliminary due to the numerosity of the observed samples, give evidence of the goodness of WI as a brain measure of cognitive load also in a clinical context, being usually used in non-clinical contexts e.g., [97,98] managing to reflect the self-reported difficulty-of-perception measure, particularly in CI users.

4.2.2. Theta

Attentional processes activate engage frontal areas involved in the generation of theta oscillations [71,99,100]. The present study results (Figure 4) showed greater pronounced frontal theta power associated with increased n-back task complexity and are reminiscent of several other studies describing theta activity in humans performing WM tasks [99,101]. Our findings could be explained by the increased attention [102–104] in correspondence to the experimental situation that was characterized by higher memory load and/or effortful cognitive processes in HC [105] and unilateral deafness children [46]. However, frontal theta activation analysis showed non-altered activity in UCI compared to controls and global a-modal processing of VWM in both groups. Moreover, it is possible to suggest that children CI users have developed adequate abilities and efficient strategies for allocating more attentional resources to support performance in a complex VWM task regardless of sensory modality, thus bridging the sensory gap due to their natural condition.

4.2.3. Gamma

Hemispheric functional specialization or functional asymmetry is a well-established characteristic of functional organization in the human brain. Verbal sound processing predominantly occurs in the left hemisphere, whereas nonverbal sound processing predominates in the right hemisphere [106]. Studies of the performance of brain-damaged children provide less clear evidence regarding functional asymmetry because such results are frequently confounded with the effects of neural plasticity [107]. Moreover, previous meta-analyses have indicated that the left prefrontal cortex (PFC) might be predominantly involved in verbal WM processes [40,108]. In previous fMRI studies, the PFC showed significant left-hemispheric lateralization during a verbal WM task and right lateralization during a spatial WM task [109].

Since the present study used auditory and visual verbal stimuli during the n-back task, and the evidence that in the processing of speech signals, binaurally presented stimuli elicit more robust brain responses in the left hemisphere [106,110], we expected that the brain would demonstrate significant left-hemispheric lateralization during task performance. This working expectation was confirmed. In fact, the almost significant difference in gamma activation between hemispheres suggests a left-hemispheric localization of VWM as proposed in the literature [40,41], regardless of deafness. Moreover, our results fit well with a previous PET study that showed that PFC activity in younger adults was left-lateralized for verbal WM stimuli [111]. So, the trend of greater gamma activation in the left hemisphere found for the VWM task independently of groups could indicate left-hemispheric lateralization of the cognitive function of auditory and visual VWM. Clearly, this is a result to be re-evaluated with a larger sample. However, this preliminary data could suggest that the cortical development of deaf children with preverbal cochlear implantation maintains the same lateralization pathway as the HC as far as the VWM. It could probably be the result of the support given by the preverbal CI that allows for maintaining brain plasticity, supporting the hypothesis of a sensitive period [112–114] for the stabilization of the integration of sensory stimuli. In fact, it is well known by EEG studies that children who received CI at an early age (<3.5 years of age at fit) showed activation of the auditory cortical areas contralateral to their cochlear implant, which resembled that of hearing subjects [115,116]. Moreover, in our data, the absence of differences in the activation of the auditory and visual area between HC and UCI argues for the lack of audio-visual cross modalities in CI users, suggesting that earlier and longer CI use would inhibit the cross-modal reorganization of auditory regions in early deafness as hypothesized by Ding and colleagues [39]. Furthermore, the comparison between HC and UCI concerning gamma activation in the left hemisphere would show that verbal WM is stronger characterized by gamma left hemisphere activation only in HC (in line with our previous work, see [28]). In fact, there were no significant differences in the other EEG frequencies in the left hemisphere between groups and within the UCI, which seem to be deficient in gamma activation during the n-back task, whereas less gamma pattern of activity is considered indicative of (inefficient) neural resource management to achieve proper cognitive performance [117]. Moreover, studies showed specific processing deviances in individuals with language and literacy problems in processing gamma rates [118,119].

The interpretation of results in terms of gamma inactivation in UCI would seem to be expressly validated during audio VWM by the activations found in the parietal area in the control group. In fact, in the parietal area, the HCs were significantly less active in the most challenging audio task than in the other conditions (Figure 5). Moreover, in addition to a strong relationship between parietal gamma activation and IES for audio conditions, we found that the first variable predicted the second (Figure 8), suggesting that gamma-band-sustained increase over parietal sites is involved with a role in WM maintenance and the binding of auditory memory representations during the n-back, in line with what was found during a different visual memory task, in the posterior—occipital areas [120,121]. These data could lead to the conclusion that gamma activation in the parietal area for auditory stimuli is the neurophysiological support for task performance only in HC children suggesting that CIs children could require greater overall recruitment of neural resources to respond similarly to the control group.

Overall, our findings confirm the role of gamma-band oscillations as candidates for the working memory function. Consistently, numerous studies observed the involvement of the gamma band in the perception and maintenance of the WM [120–122]. Moreover, functionally, gamma oscillatory activity is thought to participate in integrating neural networks within and across brain structures, facilitating coherent sensory registration [123].

Additionally, the integration of binaural input occurs via coincident counters in the superior olivary complex -SOC [124] and this brainstem region is involved in the identification of the angle and location of the sound source and the difference between the time and intensity of sounds reaching each the ear [125]. Moreover, as unilateral implant use causes abnormal reorganization of the auditory pathway at the level of the brainstem and the cortex [126], the possible key role played by SOC in achieving adaptive changes in auditory processing [127] and the evidence that SOC is the first nucleus in the central auditory pathway that receives auditory information from both ears [128], globally lead to the hypothesis that the absence in UCI of the pattern of significant parietal activation in gamma for the 2 audio condition highlighted in HC could be due precisely to the failure to achieve the processing of the binaural signal in the SOC. Their clinical condition of unilateral implanted (although all preverbal see Table 1) is in line with studies showing that monolateral deafness results in substantial changes in neural activity from the subcortical to the central auditory system [129,130] and could, in fact, result in a deficit of processing the acoustic signal and then in the non-activation of gamma-band as a result of the deficient passage of the auditory signal in the SOC. The relationship found in gamma activation with the current age and not with the auditory age nor with behavioral data (IES; ACC) could mean that the UCIs, although they have developed the brain processing that leads to the representation of the stimulus in the cortex in a similar way to the hearing person (who conversely did not show a correlation between gamma levels and age, therefore suggesting the occurred reaching of a "plateau"), would seem to assume their need for additional time to develop the same pattern of gamma activation for auditory VWM. However, this EEG pattern could be definitive due to unilateral cochlear implantation. Comparing subjects with two cochlear implants may reveal which of the two interpretations may be correct. Moreover, an assessment with auditory brainstem response (ABR) that provides information concerning the functional integrity of brainstem nuclei [131] may or may not offer support for the hypothesis of partial maturation of the SOC and thus its hypoactivation resulting in reduced gamma-band activation. However, such an analysis was not a stated purpose of the present study, but these assumptions may offer insights for further investigation.

The regression analysis conducted on the UCI group finally confirms that the chronological age predicted just parietal gamma activity for audio VWM and is not predictive for video conditions (Figure 9). Therefore, overall, the regression model seems to confirm the existence of an electroencephalographic pattern supporting VWM performance for auditory stimuli, a pattern that, however, needs to reach a certain level of brain maturation to be configured in a unilateral deaf child brain.

5. Conclusions

This is the first study investigating differences in neurophysiological patterns between hearing and UCI children during a VWM task performed in two modalities (visual and auditory). We obtained promising positive results that include the absence of differences in both behavioral performance and neurophysiological indicators of attention (frontal theta) and workload (WI) between the clinical group and controls. Although restricted to a small sample, these results can confirm the effectiveness of clinical and rehabilitation treatments for children with unilateral cochlear implants. However, the different recruitment between HC and UCI found in the parietal area for the auditory stimulation appears to be closely related to the representation of the senses at a cortical level and the different relationships found between EEG oscillations and the behavioral and biological data (current age) between HC and UCI, leading to the conclusion that the difference in activation in gamma frequency in the parietal areas can be the proper support to the auditory VWM. Thus, the synergy (absent in children with CI) between gamma activation and performance could be the reason for the extreme variability found in psychological assessments of VWM tasks in previous studies. Further studies exploring the heterogeneity of characteristics of CI users (e.g., age at implantation, comparing pre- and post-implanted groups; different etiology of onset of deafness) could provide additional support for our findings and open up future directions of investigation.

Finally, our findings, although preliminary and needing further investigations in larger samples, support evidence that EEG may hold promise in uncovering the neurophysiological mechanisms underlying the variability in VWM outcomes between HC and UCI. They may provide evidence of the activation of gamma in the parietal areas as a possible signature of neurophysiological support for auditory VWM, and to open both new lines of research on purely behavioral data, and extend existing and future rehabilitation pathways.

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Abbreviations

ABR	Auditory Brainstem Response
ACC	Accuracy
ANOVA	Analysis of Variance

AOI(s)	Area of Interest(s)
AUD	Audio
CIs	Cochlear Implants
CRs	Correct Responses
DHH	Deaf and Hard-of-Hearing
EEG	Electroencephalogram/Electroencephalography
EFs	Executive Functions
fMRI	Functional Magnetic Resonance Imaging
HC(s)	Hearing Control(s)
Hz	hertz
IAF	Individual Alpha Frequency
IES	Inverse Efficiency Score
ISI	Interstimulus Interval
m	media
ms	millisecond
PA	Parietal Area
PE	Proportion of Errors
PFC	Prefrontal Cortex
PET	Positron Emission Tomography
PSD	Power Spectral Density
RPM	Raven's standard Progressive Matrices
RTs	Reaction Times
SD	Standard Deviation
SM	Sensory Modality
SOC	Superior Olivary Complex
UCIs	Unilateral Cochlear Implant Users
VIS	Visual
WISC V	Wechsler Intelligence Scale for Children Fifth Edition
VWM	Verbal Working Memory
WI	Workload Index
WL	Mental Workload
WM	Working Memory

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Higher Right Hemisphere Gamma Band Lateralization and Suggestion of a Sensitive Period for Vocal Auditory Emotional Stimuli Recognition in Unilateral Cochlear Implant Children: An EEG Study

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In deaf children, huge emphasis was given to language; however, emotional cues decoding and production appear of pivotal importance for communication capabilities. Concerning neurophysiological correlates of emotional processing, the gamma band activity appears a useful tool adopted for emotion classification and related to the conscious elaboration of emotions. Starting from these considerations, the following items have been investigated: (i) whether emotional auditory stimuli processing differs between normal-hearing (NH) children and children using a cochlear implant (CI), given the non-physiological development of the auditory system in the latter group; (ii) whether the age at CI surgery influences emotion recognition capabilities; and (iii) in light of the right hemisphere hypothesis for emotional processing, whether the CI side influences the processing of emotional cues in unilateral CI (UCI) children. To answer these matters, 9 UCI (9.47 ± 2.33 years old) and 10 NH (10.95 ± 2.11 years old) children were asked to recognize nonverbal vocalizations belonging to three emotional states: positive (achievement, amusement, contentment, relief), negative (anger, disgust, fear, sadness), and neutral (neutral, surprise). Results showed better performances in NH than UCI children in emotional states recognition. The UCI group showed increased gamma activity lateralization index (LI) (relative higher right hemisphere activity) in comparison to the NH group in response to emotional auditory cues. Moreover, LI gamma values were negatively correlated with the percentage of correct responses in emotion recognition. Such observations could be explained by a deficit in UCI children in engaging the left

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hemisphere for more demanding emotional task, or alternatively by a higher conscious elaboration in UCI than NH children. Additionally, for the UCI group, there was no difference between the CI side and the contralateral side in gamma activity, but a higher gamma activity in the right in comparison to the left hemisphere was found. Therefore, the CI side did not appear to influence the physiologic hemispheric lateralization of emotional processing. Finally, a negative correlation was shown between the age at the CI surgery and the percentage of correct responses in emotion recognition and then suggesting the occurrence of a sensitive period for CI surgery for best emotion recognition skills development.

Keywords: lateralization index, right hemisphere emotion hypothesis, deafness, hearing loss, brain activity, length of cochlear implant use, sensitive period, auditory age

INTRODUCTION

Processing emotional expressions is fundamental for social interactions and communication; in fact, from a very young age, infants are able to detect visual and auditory information in faces and voices of people around them (Grossmann, 2010). Such capability would develop into the skill to recognize and discriminate emotions, thanks to the contribution of the experience and of the maturation of sensory and perceptual systems. This recognition involves a multisensory effect, evidenced by integration effects of facial and vocal information on cerebral activity, which are apparent both at the level of heteromodal cortical regions of convergence (e.g., bilateral posterior superior temporal sulcus), and at unimodal levels of sensory processing (Campanella and Belin, 2007; Davies-Thompson et al., 2019; Young et al., 2020).

In relation to such cross-sensorial and unisensorial effects, hearing impairment could compromise multisensory integration, in relation to its onset, etiology, and severity, leading the patient to rely only or predominantly on the visual modality in communication, including emotional perception and expression (Mildner and Koska, 2014). In fact, for 92% of children with cochlear implant (CI), perception was dominated by vision when visual and auditory speech information conflicted (Schorr et al., 2005). This statement is supported by the results of studies employing the McGurk effect on CI users, which requires the integration of auditory and visual sensory stimuli. For instance, children who received their CI prior to age 30 months accurately identified the incongruent auditory–visual stimuli, whereas children who received their CI after 30 months of age did not (Schorr, 2005). This evidence appears particularly worthy because differently from adults, who mainly prefer visual modality, infants and young children show auditory processing preference, but in children with congenital hearing impairment, such auditory dominance appears absent. Interestingly, in post-lingually deaf CI patients, such greater relying on visual information, indexed by higher speech-reading performances than normal-hearing (NH) individuals, led instead to an increased capacity of integrating visual and distorted speech signals, producing higher visuoauditory performances (Rouger et al., 2007). Furthermore, such evidence in post-lingual deaf patients was also supported by neurophysiological assessments, evidencing a positive correlation between visual activity and auditory speech recovery, suggesting a facilitating role for the visual modality in auditory words' perception during communicative situations (Strelnikov et al., 2013). With respect to general processing preferences, contrary to adults, who prefer the visual modality (Scherer, 2003), infants and young children exhibit auditory processing preference. Importantly, congenital hearing-impaired children who underwent auditory–verbal therapy (a therapy limiting visual cue in order to strengthen the auditory pathway for language learning) reported a behavior similar to NH children, which is an overall auditory preference in response to audiovisual stimuli, although responses did not significantly differ from chance (Zupan and Sussman, 2009). Contrary to NH individuals, those with hearing impairments do not benefit from the addition of the auditory cues to the visual mode (e.g., Most and Aviner, 2009). Although the accuracy of emotion perception among children with hearing loss (HL) was lower than that of NH children in auditory, visual, and auditory–visual conditions, in prelingually deaf very young children (about 4–6 years old), the combined auditory–visual mode significantly surpassed the auditory or visual modes alone, as in the NH group, supporting the use of auditory information for emotion perception, probably thanks to intensive rehabilitation (Most and Michaelis, 2012) and neuroplasticity. Such results strongly support the hypothesis of a sensitive period (Kral et al., 2001; Sharma et al., 2005; Gilley et al., 2010) for the establishment of the integration of auditory and visual stimuli.

Thanks to their activity of direct stimulation of the acoustic nerve, converting the auditory stimuli into electrical signals directed to the brain, CIs can successfully restore hearing in profoundly deaf individuals. After intensive rehabilitation, most CI users can reach a good level of speech comprehension. However, the acoustic signal provided by the device is severely degraded, resulting in a poor frequency resolution and deficits in pitch patterns (Gfeller et al., 2007; eHopyan et al., 2012) and pitch changes or direction discrimination (Gfeller et al., 2002) in comparison to NH controls.

Hearing-impaired children go through an early auditory development that is different from that of NH toddlers. This condition would affect their judgment of the emotional content of a stimulus, insofar as the auditory modality resulted as particularly important for the communication of emotions in young children (Baldwin and Moses, 1996; Akhtar and Gernsbacher, 2008). The study of such mechanisms appears of great impact since about 600,000 patients world-wide are CI users (The Ear Foundation, 2017), and many of them are children who were born deaf or lost their hearing within the first few years of life. CI children are a paradigmatic model for the study of emotion recognition skills, as due to the early acquisition of deafness, they learned language through the degraded input of the CI, which greatly affects harmonic pitch perception. This ability is strongly necessary for emotion recognition in voices, and its deficiency could have implications on how child CI users learn to produce vocal emotions (Damm et al., 2019). However, a very recent study provided evidence that also deaf people can develop skills for emotional vocalizations despite the presence of some differences in comparison to NH adults (Sauter et al., 2019). Using unilateral CI (UCI) in children, due to non-physiological development of their auditory system and to their asymmetry in receiving auditory inputs, represents a powerful model of investigation of the possible modulation of the hemispheric specialization and of auditory-related emotional skills development in relation to the restored hearing condition. Additionally, such participants would provide evidence of the possible modulation of the physiological processes of emotion recognition following the restoration of the auditory capabilities, of which the exact time of beginning is due to the CI surgery time. Children, 7–13 years of age, using UCIs perform more poorly than age- and gender-matched controls on the affective speech prosody task but as well as controls in tasks of facial affect perception (Hopyan-Misakyan et al., 2009), as measured by the DANVA-2 (Nowicki and Duke, 1994).

One of the few studies that investigated both auditory recognition and vocal production of emotions did not find any consistent advantage for age-matched NH participants in comparison to three prelingually, bilaterally, profoundly deaf children aged 6–7 years who received CIs before age 2 years; however, confusion matrices among three of the investigated emotions (anger, happiness, and fear) showed that children with and without hearing impairment may rely on different cues (Mildner and Koska, 2014).

With respect to emotional skills attainment and in relation to the hemispheric specialization for emotional processing (Gainotti, 2019), it is interesting to consider that patients enrolled in the present study were UCI users, that is, single-side deaf (SSD) patients. In fact, in SSD population, it was evidenced that the occurrence of a massive reorganization of aural preference in favor of the hearing ear is greater than the precocity of unilateral HL onset, therefore supporting the importance of a short time between the first and second implantation in children (Kral et al., 2013; Gordon et al., 2015; Gordon and Papsin, 2019).

Concerning neural correlates of emotion recognition, gamma band electroencephalogram (EEG) was found to be particularly sensitive for emotion classification (Li and Lu, 2009; Yang et al., 2020). Gamma band cerebral activity has been previously linked to facial emotion recognition processes; for instance, a right hemisphere dominance in gamma activity was found during emotional processing of faces in comparison to neutral ones (e.g., Balconi and Lucchiari, 2008). Such evidences are in accord to the right hemisphere hypothesis for emotion processing, that starting from observations on patients with single hemisphere lesions states the dominance of the right hemisphere for every kind of emotional response (Gainotti, 2019). With specific regard to emotional prosody processing and brain activity lateralization, Kotz and colleagues hypothesized that (i) differentially lateralized subprocesses underlie emotional prosody processing and (ii) the lateralization of emotional prosody can be modulated by methodological factors (Kotz et al., 2006). Furthermore, concerning verbal stimuli, in adult CI users, gamma band–induced activity was found to be higher in NH than in CI users, irrespectively of the valence of the emotions investigated (Agrawal et al., 2013).

On the base of the previous issues, the following experimental questions have been approached in a population of NH and UCI children: (i) Given the non-physiological development of the auditory system in deaf children who underwent hearing restoration through CI use, are the emotional auditory stimuli processed in a similar way than NH children? (ii) Is the auditory age, meant as the age at CI surgery, crucial in the capacity of recognizing emotions? (iii) In light of the evidence that the right hemisphere has a unique contribution in emotional processing – summarized in the right hemisphere emotion hypothesis – does the side of the CI influence the processing of emotional cues in UCI children, or is the "physiological right lateralization" respected?

MATERIALS AND METHODS

Participants

For the present study, 10 NH (6 female, 4 male; 10.95 ± 2.11 years old) and 9 UCI user (UCI; 5 female, 4 male; 9.47 ± 2.33 years old) children were enrolled. Six children had their CI in their right ear and three in their left ear; at the moment of the test, none of them wore any hearing aid in their contralateral ear. All participants were right-handed except for two children: one belonging to the NH and one to the UCI group. Further clinical details of the UCI group are reported in **Table 1**.

Protocol

The task consisted of the recognition of nonverbal vocalizations belonging to a database previously validated and employed in several studies (Sauter et al., 2006, 2010, 2013) and grouped into three emotional states: positive (achievement, amusement, contentment, relief), negative (anger, disgust, fear, sadness), and neutral (neutral, surprise), which participants were asked to match with the corresponding emotional picture (**Figure 1**). For each emotion, six different audio stimuli were reproduced, whereas there was a single corresponding emotional picture for each emotion. The emotional audio stimuli had a mean duration of 1,354.25 ± 223.39 ms and were delivered at 65 dB HL (Cartocci et al., 2015, 2018; Marsella et al., 2017; Piccioni et al., 2018) through two loudspeakers placed in front of and behind the participant at the distance of 1 m each, to meet CIs' best requirements for their use. Participants underwent training with the kind of emotional stimuli employed in the study

TABLE 1 | Demographics concerning the UCI group, in particular etiology of deafness, its onset, and duration of deafness before CI surgery.

Participants	Age (years)	Etiology	Onset of deafness	Period of Deafness (years)
PI	11,39	Unknown	Birth	1,38
P2	12,04	Unknown	3 years old	5,91
P3	11,66	Unknown	4 years old	2,25
P4	10,22	Homozygous mutation of the connexin-26 gene	Birth	1,11
P5	7,08	Congenital CMV infection	Birth	3,82
P6	9,99	Homozygous mutation of the connexin-26 gene	Birth	2,93
P7	9,24	Homozygous mutation of the connexin-26 gene	Birth	8,16
P8	12,57	Unknown	3,5 years old	6,41
P9	14,37	Unknown	Birth	13,18

and a familiarization with the experimental protocol. Once the researcher verified the comprehension of the emotional stimuli and the task by the participant, he/she was asked to carefully listen to the emotional audio and then to identify the emotion reproduced by the stimulus pressing one out of five buttons on a customized keyboard, corresponding to the target emotional picture. For instance, the participant heard a laugh, and he/she had to identify the corresponding picture, a smiling young lady, out of five options. There was no time limit set for such identification and matching with the target emotion. Each picture representing the target emotion was placed at least once (and maximum twice) in each of the five positions on the screen. The number of five pictures among which the participant had to identify the target stimulus was chosen in accordance with Orsini et al. (1987), who found for the range of age of the enrolled participants a digit span of more than 4.5 items for both males and females. Stimuli were delivered through E-prime software, in a pseudorandomized order so that it was not possible that two stimuli belonging to the same emotion were consecutive.

The study was carefully explained to all participants and to their parents, who signed an informed consent to the participation. The study was approved by the Bambino Gesù Pediatric Hospital Ethic Committee, protocol 705/FS, and was conducted according to the principles outlined in the Declaration of Helsinki of 1975, as revised in 2000.

EEG

A digital EEG system (BE plus EBNeuro, Italy) was used to record 16 EEG channels (Fp, Fz, F3, F4, F7, F8, T7, T8, P3, P4, P7, P8, O1, O2) according to the international 10/20 system, with a sampling frequency of 256 Hz. The impedances were maintained below 10 kΩ, and a 50-Hz notch filter was applied to remove the power interference. A ground electrode was placed on the forehead and reference electrodes on earlobes. The EEG signal was initially bandpass filtered with a fifth-order Butterworth filter (high-pass filter: cutoff frequency fc = 1 Hz; low-pass filter: cutoff frequency fc = 40 Hz). Because we could not apply independent component analysis because of the low number of EEG channels (i.e., 16), we used a regression-based method to identify and correct eye-blinks artifacts. In particular, the Fpz channel was used to identify and remove eye-blink artifacts by the REBLINCA algorithm (Di Flumeri et al., 2016). This method allows the EEG signal to be corrected without losing data. For other sources of artifacts (e.g., environmental noise, user movements, etc.), specific procedures of the EEGLAB toolbox were employed (Delorme and Makeig, 2004). In particular, the EEG dataset was first segmented into epochs of 2 s through moving windows shifted by 0.125 s. This windowing was chosen with the compromise of having a high number of observations, in comparison with the number of variables, and in order to respect the condition of stationarity of the EEG signal. This is in fact a necessary assumption in order to proceed with the spectral analysis of the signal. Successively, three criteria were applied to those EEG epochs (Aricò et al., 2017; Borghini et al., 2017): (i) threshold criterion (amplitudes exceeding ± 100 μV); (ii) trend criterion (slope higher than 10 μV/s); and (iii) sample-to-sample criterion (sample-to-sample amplitude difference >25 μV).

All EEG epochs marked as "artifact" were removed in order to have a clean EEG signal. In order to accurately define EEG bands of interest, for each participant the individual alpha frequency (IAF) was computed on a closed-eyes segment recorded prior to the experimental task. Thus, the EEG was filtered in the following frequency bands: theta [IAF − 6 ÷ IAF − 2 Hz], alpha [IAF − 2 ÷ IAF + 2 Hz], beta [IAF + 2 ÷ IAF + 16 Hz], and gamma [IAF + 16 ÷ IAF + 25 Hz] (Klimesch, 1999). EEG recordings were segmented into trials, corresponding to audio stimulus listening and target picture matching. The power spectrum density was calculated in correspondence of the different conditions with a frequency resolution of 0.5 Hz. Trials were normalized by subtracting the open-eyes activity recorded before the beginning of the experimental task.

Lateralization Index

The lateralization index (LI) was calculated in order to assess the relative asymmetry between the two cerebral hemispheres' activity during the task execution (audio stimuli perception and target visual stimuli matching), as the right hemisphere theory for emotion predicts a relative higher right activation during emotional stimuli processing.

The LI was calculated on the basis of the formula previously adopted by Vanvooren et al. (2015):

$$\mathbf{L}I = \frac{\mathbf{R-S}}{\mathbf{RS}}$$

where R stands for right hemisphere, and L for left hemisphere. The LI ranges from +1, for cortical activity entirely asymmetrical

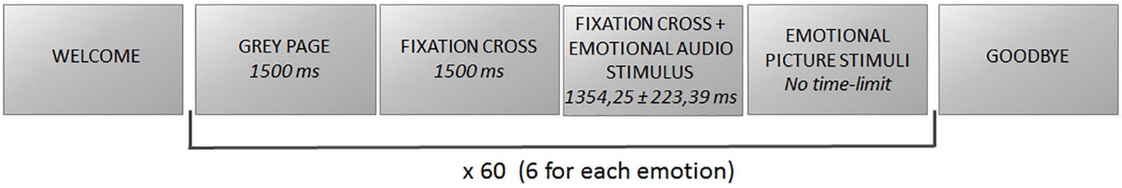

FIGURE 1 | Scheme of the experimental protocol.

to the right hemisphere, to zero for symmetrical cortical activity, and −1 for cortical activity entirely asymmetrical to the left hemisphere. For the right hemisphere activity calculation, the estimation from the following electrodes was averaged: F4, F8, T8, P4, P8, O2, whereas for the left hemisphere. It was averaged from the following ones: F3, F7, T7, P3, P7, O1. The LI was already employed on hearing-impaired children, in particular, SSD children, finding an asymmetry in cortical activity during the execution of a word in noise recognition task influenced by the direction of the background noise in SSD but not in NH children (Cartocci et al., 2019).

Statistical Analysis

Both the percentage of correct responses and LI data were compared between the NH and UCI groups through analysis of variance (ANOVA) with two factors: GROUP (2 levels: NH and UCI) and EMOTIONAL STATE (3 levels: positive, negative, and neutral). A simple regression analysis was performed for investigating the relation between (i) the percentage of correct responses and the LI values, (ii) between the percentage of correct responses and the age at the test execution, and (iii) between the percentage of correct responses and the age at CI surgery.

RESULTS

Behavioral results evidenced a higher percentage of correct responses provided by NH children in comparison to UCI children ($F = 18.898$, $p < 0.001$, partial $\eta^2 = 0.270$) (**Table 2**), but an effect of the emotional state was not seen ($F = 1.890$, $p = 0.161$, partial $\eta^2 = 0.069$), although for both groups the neutral cues were the most difficult to recognize. Neither the interaction between the variable group and emotional state ($F = 0.032$, $p = 0.968$, partial $\eta^2 = 0.001$) was observed (**Figure 2**).

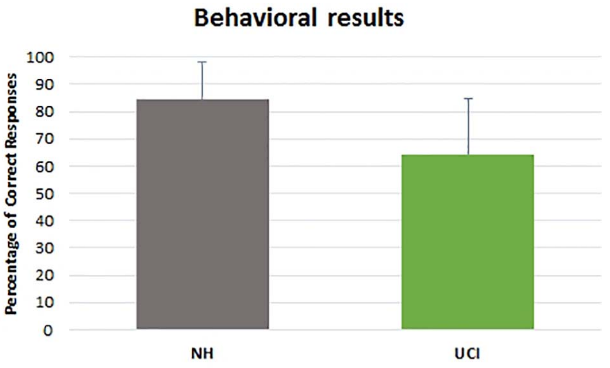

FIGURE 2 | Percentage of correct responses reported by NH and UCI children. Bars describe means, and error bars describe standard deviations.

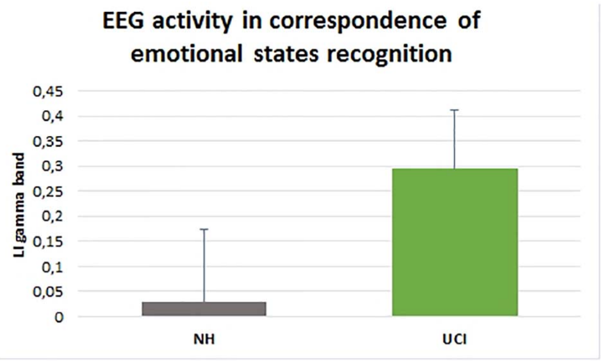

FIGURE 3 | Gamma band activity LI in NH and UCI children. Bars describe means, and error bars describe standard deviations.

ANOVA results showed higher LI values, indicating a higher activity in gamma band in the right in comparison to the left hemisphere, in UCI in comparison to NH children ($F = 58.656$, $p < 0.001$, partial $\eta^2 = 0.535$) (**Figure 3**), irrespectively of the emotional state (negative, neutral, and positive) ($F = 1.686$, $p = 0.195$, partial $\eta^2 = 0.062$). Additionally, any interaction between the variable groups and emotional state was not found ($F = 1.121$, $p = 0.333$, partial $\eta^2 = 0.042$).

TABLE 2 | Mean percentages of correct responses ± standard deviation for each group (UCI and NH) and for each emotional state.

Group	Negative	Neutral	Positive
NH	86,58% ± 9,82	78,33% ± 18,92	88,33% ± 10,17
UCI	65,05% ± 19,37	58,24% ± 22,17	69,67% ± 21,02

A negative correlation was observed between LI gamma values and the percentage of correct responses ($F = 11.801$, $p = 0.001$, $r = -0.420$, partial $\eta^2 = 0.177$) (**Figure 4**).

Additionally, for the UCI group, any difference between the CI side and the deaf contralateral side in the gamma activity was not

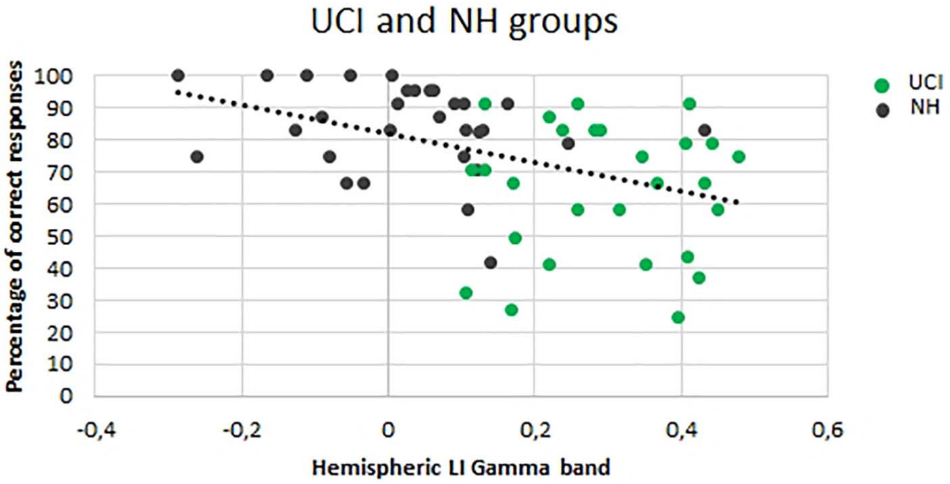

FIGURE 4 | Correlation between the lateralization (LI) gamma values and percentage of correct responses for both the UCI and NH groups. Dark dots represent NH values, and green dots represent UCI values.

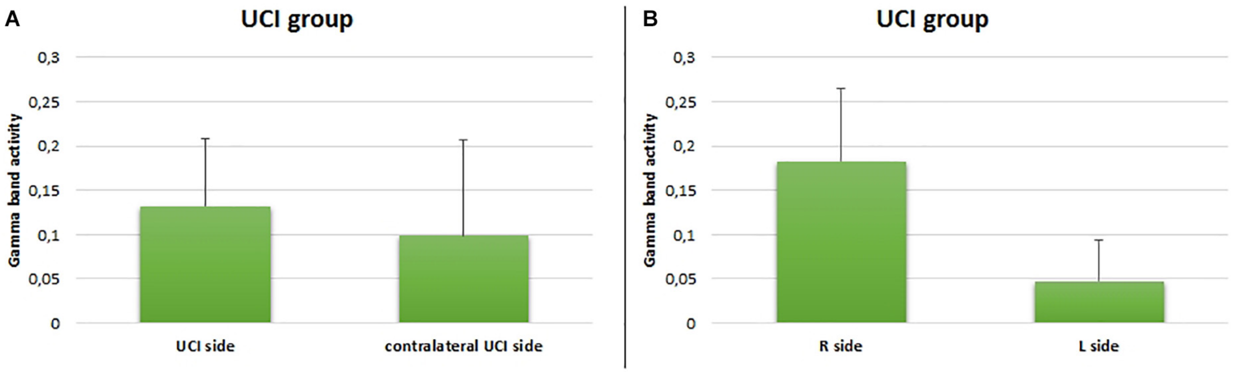

FIGURE 5 | Comparison between gamma activity in the UCI group with respect to the UCI side **(A)** and right or left side **(B)**. Bars describe means, and error bars describe standard deviations.

shown ($F = 0.598$, $p = 0.212$, partial $\eta^2 = 0.032$) (**Figure 5A**), but a higher gamma activity in the right in comparison to the left hemisphere was found ($F = 54.552$, $p < 0.001$, partial $\eta^2 = 0.532$) (**Figure 5B**).

For the UCI group, no correlation was found between the age of the UCI children at the moment of the experiment and the percentage of correct responses ($F = 0.052$, $p = 0.821$, $r = 0.046$, partial $\eta^2 = 0.002$), similarly to the NH children group ($F = 1.130$, $p = 0.297$, $r = 0.197$, partial $\eta^2 = 0.039$). Additionally, a negative correlation was shown between the age at the CI surgery and the percentage of correct response reported by UCI children ($F = 7.030$, $p = 0.014$, $r = 0.468$, partial $\eta^2 = 0.219$) (**Figure 6**). Finally, when calculating the mean of the correct responses for each participant, irrespective of the emotional states, despite the lack of significance ($F = 3.056$, $p = 0.124$, $r = -0.551$, partial $\eta^2 = 0.304$), a higher percentage of correct responses was highlighted, higher than 70%, only in early implanted children, that is, before 3.5 years of age (**Figure 6**, black dots).

DISCUSSION

According to literature, the lower percentage of correct responses provided by UCI children in comparison to NH children highlights their impairment in vocal emotion recognition skills (Agrawal et al., 2013; Wiefferink et al., 2013; Chatterjee et al., 2015; Jiam et al., 2017; Ahmed et al., 2018; Paquette et al., 2018). This would be strongly related to the preverbal and periverbal deafness acquisition. In fact, in a study employing emotional vocal stimuli in adult CI users, such performance difference was not shown (Deroche et al., 2019). Furthermore, there are evidences of different strategies implemented by CI and NH listeners for emotional stimuli recognition, more based on pitch range cues in the former and more relying on mean pitch in the latter group (Gilbers et al., 2015). In addition, such deficit in emotion recognition in UCI children in comparison to NH children appears strictly related to the matter of social interaction and social development (Jiam et al., 2017); in fact, a correlation between impairments in perception and production of voice

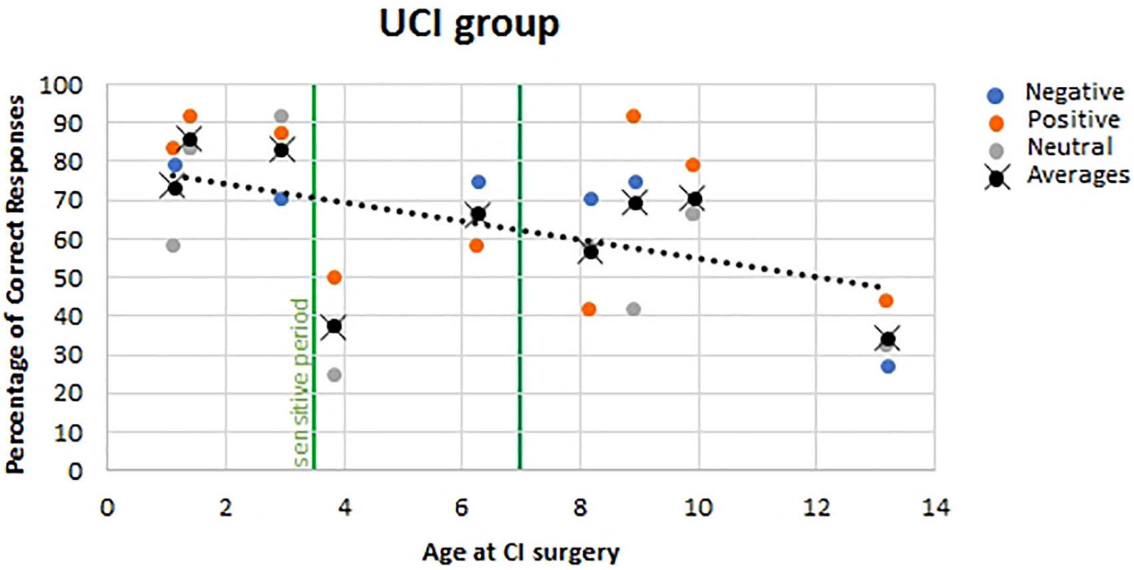

FIGURE 6 | Correlation between age at CI surgery and percentage of correct responses in UCI children. Orange dots stand for positive emotional states; blue dots stand for negative emotional states, and gray dots stand for neutral emotional states. Black dots stand for the mean of correct responses for each participant, irrespective of the emotional state. The vertical green lines represent the sensitive period threshold (3.5 and 7 years old) for the central auditory system development (Sharma et al., 2005).

emotion was found, like in the case of infant-directed speech, and in 5- to 13-year-old children who used CI (Nakata et al., 2012). It is interesting to note that a previous study employing vocal child-directed happy and sad speech stimuli reported higher performance in NH in comparison to CI using children; however, the percentage of recognition was higher than the one reported in the present study, probably due to the child-directed characteristic of the stimuli (Volkova et al., 2013).

Concerning the difference in gamma LI values observed in UCI in comparison to the NH group, it confirmed a difference in gamma band activity previously reported by Agrawal et al. (2013) in comparison between the same groups, therefore supporting the suitability of the study of gamma rhythms in the investigation of emotional messages conveyed by means of auditory stimuli. However, the previously mentioned study and the present study are not perfectly comparable because of the differences (i) in the sample – adults and children, respectively, – and therefore plausibly in the etiology of deafness; (ii) in the location of EEG activity acquisition, that is, Cz and multiple electrodes over the two hemispheres, respectively; and (iii) in the kind of emotional stimuli, that is, verbal stimuli pronounced with neutral, happy, and angry prosody in Agrawal and colleagues' study, while vocal nonverbal stimuli belonging to 10 emotions grouped into three emotional states in the present study. Moreover, the higher LI values reported for UCI in comparison to NH children would imply a more sustained conscious processing of the stimuli for the NH group in comparison to the UCI group and a higher processing of the emotional face stimuli – employed for the matching of the auditory stimuli for the identification of the target emotion – by the UCI group (Balconi and Lucchiari, 2008). In fact, McGurk studies showed a higher relying of UCI children on the visual sensation than on the auditory one in case of uncertainty (Schorr et al., 2005).

The correlation between higher right lateralization, as indexed by higher LI values, and the percentage of correct responses could be explained by the evidence of higher activation and asymmetry levels in poorer performers in emotion-in-voice recognition tasks than those of more proficient ones (Kislova and Rusalova, 2009). This possibly also reflects the poorer performance in emotion recognition obtained by UCI children, as well as their higher LI values in comparison to NH children. In fact, it was shown by studies on single hemisphere damage that although the right hemisphere is responsible for low-level discrimination and recognition of affective prosody, in case of higher task demands in terms of associational-cognitive requirements, the left hemisphere is engaged (Tompkins and Flowers, 1985). Thus, UCI children would present deficits in such engaging of the left hemisphere for more complex emotional processing tasks. This could be explained by the neuroimaging evidence that indeed areas appearing to be primarily involved in emotional prosodic processing, that is, posterior temporal (parietal) brain regions (Kotz et al., 2006), are the same areas presumably more involved by the neuroplastic changes that occurred after CI surgery (Giraud et al., 2000; Kang et al., 2004) and the following hearing sensation restoration.

The negative correlation between age of implantation and percentage of correct responses in emotion recognition is in accordance with previous studies (Mancini et al., 2016). On the contrary, in the Deroche and colleagues' study on adult CI users cited above, any effect of the age at implantation on the emotion recognition was not found, but this would be caused by the post-lingual acquisition of deafness in the majority of the sample (19 over 22 CI users) and by the type of emotions investigated, which is happy, sad, and neutral, whereas in the present study, 10 emotions were employed (Deroche et al., 2019). Furthermore, in Volkova et al.' (2013) study, employing child-directed emotional speech, performance of the children CI users was positively associated with duration of implant use. Such evidence could be compared to present results, given the almost overlap between age at CI surgery and length of CI use in the enrolled sample. In addition, the trend that better performances were obtained by children implanted before 3.5 years old suggests the influence of a sensitive period, identified through P1 cortical auditory-evoked potential trajectory post-CI development (Sharma et al., 2002, 2005; Sharma and Dorman, 2006; Kral and Sharma, 2012; Kral et al., 2019) also on emotion recognition skills development. Such phenomenon could be explained by the better auditory–visual integration achieved by children implanted before 3.5 years of age as shown by Miller's test of the race model inequality executed by early and late implanted children (Gilley et al., 2010). Such auditory–visual integration capability achievement is also witnessed by McGurk effect tests on CI children, showing that 38% of early implanted children – before the age of 2.5 years – but none of the late implanted children exhibited the bimodal fusion occurring in the McGurk effect, being instead biased toward the visual modality in contrast to the NH children who were biased toward the audio modality (Schorr et al., 2005). These evidences, with respect to the topic of emotion recognition skills development, are in accord to studies indicating that auditory and visual integration is necessary for the achievement of such capabilities (Campanella and Belin, 2007). In relation to this matter, there is also the evidence of a delay on facial emotion recognition in preschoolers using CI (and hearing aids) in comparison to NH mates, and interestingly, there was not any correlation between facial emotion recognition and language abilities (Wang et al., 2011). Differently, another study found a relation between better language skills and higher social competence, both in NH and CI children, although in the latter group, less adequate emotion-regulation strategies and less social competence than NH children were highlighted (Wiefferink et al., 2012). In addition, a study investigating both linguistic (recognition of monosyllabic words and of key words from sentences within background noise; repetition of non-words) and indexical (discrimination of across-gender and within-gender talkers; identification of emotional content from spoken sentences) properties in perceptual analysis of speech in CI children found an association between better performances in such feature recognition and a younger age at implantation (and use of more novel speech processor technology) (Geers et al., 2013).

Moreover, concerning the emotional communication, a suggestion of deficits also in the imitation of emotional (happy and sad) speech stimuli was found (Wang et al., 2013). Therefore, it sharply results in the vision and need of two targets of rehabilitation for children with CI that should be treated both conjointly and separately: language treatment and emotional intervention.

CONCLUSION

In light of the present results, in relation to the experimental questions previously declared, it is possible to conclude that (i) the processing of the emotional stimuli by deaf children using CI appears to be different from NH children, as suggested by the higher relative right hemisphere gamma band activity, possibly explained by the non-physiological development of the auditory system; (ii) on account of the inverse correlation between the age at the CI surgery and the percentage of correct responses, the precocity of performing the CI surgery for the attainment of best emotion recognition skills appears crucial, probably because of neuroplastic changes allowing a better processing and categorization of emotional stimuli; and (iii) the CI side does not appear to influence the processing of emotional stimuli, although interestingly the relative higher gamma band activity appears to be counterproductive in terms of emotion recognition performances; such aspect needs further investigation at the light of the possible particular implications of the right hemisphere hypothesis (Kotz et al., 2006).

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Bambino Gesù Pediatric Hospital Ethics Committee. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

GC conceived and conducted the study, performed the data analysis, and wrote the manuscript. AG and BI prepared the experimental protocol, conducted the study, and elaborated data. AS, SGi, AD, SGa, RG, CL, PL, and FF enrolled patients and organized experimental sessions. PM provided support for the organization and realization of the study. AS and FB edited the manuscript. FB supervised the entire experiment. All authors read and approved the final version of the article.

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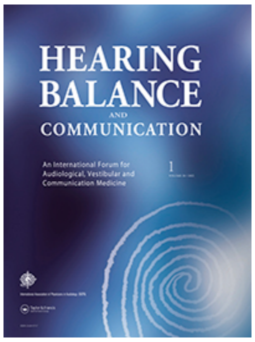

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‘Musical effort’ and ‘musical pleasantness’: a pilot study on the neurophysiological correlates of classical music listening in adults normal hearing and unilateral cochlear implant users

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ORIGINAL ARTICLE

'Musical effort' and 'musical pleasantness': a pilot study on the neurophysiological correlates of classical music listening in adults normal hearing and unilateral cochlear implant users

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ABSTRACT

Objective: This pilot case-control study is aimed to estimate 'musical effort' and 'musical pleasantness' in adults normal hearing (NH) and Unilateral Cochlear Implant (UCI) users via neurophysiological indices and a cognitive-behavioural approach.

Methods: 7 NH and 7 UCI subjects underwent electroencephalographic (EEG) recording with two EEG indices, Approach Withdrawal (AW) and Workload (WI), and behavioural tests conducted during a forced-choice musical emotion recognition task performed in quiet and noise in two emotional conditions.

Results: Musical effort in terms of WI did not vary between conditions and groups. Inversely, for AW, statistically significant differences in noisy conditions were observed between groups, indicating a difficulty for the patients in discriminating the musical pleasantness in noise. However, self-reported data suggest the ability of UCI to cognitively evaluate the stimulus in terms of both auditory difficulty and pleasantness.

Conclusion: Findings suggest the possible relevance of the Cochlear Implant in supporting the patients' effort in listening to music in noise, allowing a pleasant enjoyment in the quiet condition. Overall, this study appears worthy of interest and paves the way for further investigation by providing data on the neurophysiological perception of music.

KEYWORDS
Music; hearing loss; electroencephalography; pleasantness; effort

1. Introduction

'Our auditory systems, our nervous systems, are tuned for music. Perhaps we are a musical species no less than a linguistic one'. With this masterful hypothesis put forward by Oliver Sacks [1], it is interesting to dialogue reflecting on how real this can be for people with a bionic auditory system, such as cochlear implant users.

Both music and language use modulation of acoustic parameters to transduce information [2] conveying important affective and expressive messages. However, music, especially without lyrics [3], is a more complex auditory stimulus than language, requiring more advanced resolution and comprehension mechanisms [4,5].

While the restoration of speech perception in CI users has reached very high technical standard levels [6–9], the perception and enjoyment of music through Cochlear Implant (CI) is still an open challenge [10]. In fact, although CI users rank music as the second most important acoustic stimulus in their lives next to understanding speech [11], even many years after the auditory restoration, they report difficulties in music perception [12,13]. Indeed, even though structural features of music (i.e. rhythmic patterns) are effectively perceived with similar accuracy as normal hearing people (NH) [10,14] the perception of other essential features (such as pitch discrimination, melody and timbre recognition) and speech perception in noise are still extremely challenging for

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CIs [12,15,16]. Despite the impaired pitch perception, literature findings suggest that CI users might recognise emotional valence conveyed by music [17]. Moreover, impairments in music perception in adults with CI are often reported as significant negative factors in self-reports of quality of life [18].

Listening to music is amongst the most rewarding experiences for humans [19] and often people listening to music for pleasure [20]; so, adults CI users are very disappointed that they can no longer hear and appreciate music after receiving their implant [18] reporting a decreased enjoyment in listening to music following [21–23]. Moreover, they tend to rate the tone quality as less pleasant than do NH adults [24–26] and to perceive more auditory fatigue during listening to music [27].

Surprisingly, research on 'musical effort' – the broad process of allocation of attentional resources during music performance or listening [28,29] – has been so far an understudied aspect of music cognition [30], while extensive studies investigate effort in speech listening through different techniques such as self-report [31], behavioural [32] and neurophysiological [33–35].Moreover, no study to date has examined electroencephalographic (EEG) correlates of musical effort in cochlear implanted patients.

The pre-mentioned evidences and the fact that as highlighted by Faulkner and Pisoni [18] music perception is not routinely assessed clinically by audiologists primarily interested in speech-language recognition outcomes (see [5,36,37] for music perception assessment in CIs) offer auditory neuroscience [38] the opportunity to investigate through the EEG, a direct, non-invasive and widely used tool [39,40] with excellent temporal and good spatial resolution, the cortical correlates of music perception to improve the clinical assessment of the patient and his/her quality of life.

Therefore, the aim of this pilot study is to investigate pleasantness and effort in classical music perception under different listening conditions in CI users and in normal hearing adults through neurophysiological indexes and cognitive-behavioural measures.

2. Materials and methods

2.1. Participants

7 unilateral cochlear implant (UCI) adults have been recruited and personal and clinical data are summarised in Table 1. On the day of the EEG registration, all patients previously underwent warble -tone -free field and speech audiometry to ensure their hearing abilities were adequate for the task and they were not aided in the other ear. Word recognition of at least 50% at 65 dB SPL from Italian Speech Audiometry [41] was the inclusion criterium for CI users. The NH control group was composed of 7 participants (4 F;3M; 44.142yrs old). The study was conducted in accordance with the Declaration of Helsinki of and was formally approved by the competent ethics committee. All participants were not musicians and provided an informed written consent before beginning the experimental protocol.

Table 1. Patient personal and clinical data.

Patient	Deafness	Gender	Age (yrs)	Auditory Age (mos)	Side of CI
1	Prelingual	F	40	36	L
2	Prelingual	F	22	9	R
3	Postlingual	F	68	108	L
4	Postlingual	F	58	10	R
5	Prelingual	M	41	111	R
6	Postlingual	F	50	12	L
7	Prelingual	M	61	108	R

2.2. Stimuli

Extracts of western classical instrumental music, originally not composed for singing voice, drawn from different periods (Baroque e.g. J.S. Bach St Matthew Passion; Classical e.g. L. van Beethoven Symphony 3; Contemporary e.g. Grieg Peer Gynt) present in a validated database [42–44], were used. Importantly, although the original instrumentation was generally orchestral or for piano, the extracts all consisted of transcriptions for 'piano solo' to simplify the complexity of the multi-instrumental composition which could create perceptual bias due to the difficulties of CI users in processing instrumental timbres [45,46]. In addition, to avoid interpretative bias on EEG data, the transcription for a percussion instrument such as the piano made possible to simplify and standardise perception for CI users who show greater ability to identify percussive than woodwind or brass instruments [15]. From the 32 database stimuli categorised according to the basic characterising emotion (happiness-H, sadness-S) 16 were selected. Then, to create different listening conditions, each musical excerpt was processed using a continuous 4 talker babble background noise as the competitive signal to provide informational masking by creating 3 versions based on different levels of signal/Noise ratio (SNR) [47] as adopted in previous EEG studies [35,48]. The 3 listening conditions were: quiet (Q), a moderate SNR + 10 (X) and a more challenging SNR + 5 (V). Finally, two stimuli lists were created, each of 8 musical excerpts in the respective 3 listening conditions. The lists were randomised among the 14 participants, 24 stimuli from each list were randomly presented with a different sequence for each participant.

2.3. Experimental protocol

Subjects performed a musical emotion-forced choice recognition task in 3 listening conditions with simultaneous EEG recording, comfortably seated on a chair in an electrically-shielded room in front of the stimulating PC. Stimuli were transmitted through two loudspeakers placed in front of and behind the participant's head at a distance of 1 m each, to meet CIs' best requirements for their use. Each subject was asked to listen carefully to the musical excerpts, which were preceded by a white screen (1500 ms) and a fixation cross (3000 ms). At the end of each stimulus, two emoticons appeared on the screen indicating a sad and a happy face [49] and the subject had to indicate, using two specific buttons on the keyboard, whether the music listened to express a sad or happy emotion. Subsequently, stylised icons appeared on the screen through which the participant had to indicate with the mouse the level of difficulty perceived in listening (*easy*, *medium*, *difficult*) and his subjective level of pleasantness (*I like it*, *Indifferent*, *I do not like it*) (Figure 1 for protocol schematisation). Before starting, each subject was given detailed information about the task and was made familiar with stimuli similar though different from the one used in the test. The entire protocol was built and controlled by E-Prime (Psychology Software Tools, Pittsburgh, Pa Version 3.0) software which was also used for the acquisition of the cognitive and behavioural data.

2.4. Behavioural data

Behavioural measures in terms of Reaction Times (RTs) – ms taken by the participant to categorise the emotion of the stimulusand accuracy (ACC) in terms of the percentage of correct responses–were recorded. In addition, to capture the cognitive component of perception, ratings in terms of difficulty and pleasantness given by each participant were collected.

2.5. EEG recording and signal processing

An EEG system (BEPlus, EBNeuro spa, Italy) at a sampling rate of 256 Hz was used to record electrical potentials by means of an electrode cap, according to the 10–20 international system [50]. In this case the following 20 channels were considered for analysis (already considered in experimental protocols with NH and HI subjects [39]: Fpz, Fz, F3, F4, F7, F8, Cz, C3, C4, Cp5, Cp6, T7, T8, Pz, P3, P4, P7, P8, O1, O2. The impedances were maintained below 10 kΩ, and a 50-Hz notch filter was applied to remove the power interference. The ground electrode was placed on the forehead and the reference electrodes on the earlobes. The EEG signal has been band pass filter at 1–40 Hz with a 5th-order Butterworth filter. To identify and correct blink artefacts, a regression-method was used without losing data. More specifically, the REBLINCA algorithm [51] allowed the identification and removal of eye-blink artefacts. Moreover, specific procedures of the EEGLAB toolbox [52] were employed to remove other sources of artefacts. With the intention of obtaining a high number of observations, in comparison with the number of variables, and in order to respect the condition of stationarity of the EEG signal (a necessary assumption in order to proceed with the spectral analysis of the signal), the EEG trace was

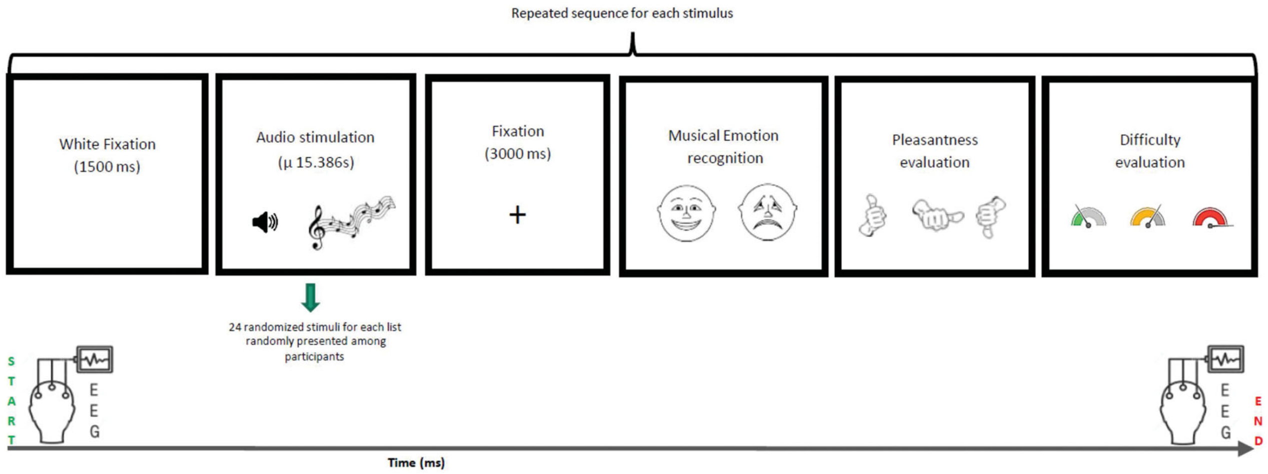

Figure 1. Schematisation of the experimental protocol.

segmented into epochs of 2 s through moving windows shifted by 0.125 s. Successively, three criteria were applied to those EEG epochs [53,54]: (i) threshold criterion (amplitudes exceeding ± 100 μV); (ii) trend criterion (slope higher than 10 μV/s); and (iii) sample-to-sample criterion (sample-to-sample amplitude difference >25 μV). All EEG epochs marked as 'artifact' were removed to have a clean EEG signal. Subsequently, Individual Alpha frequency (IAF) was computed for each subject on a closed-eyes segment recorded prior to the experimental task to define EEG bands of interest. Thus, according to the scientific literature [55] the EEG was filtered in the following frequency bands: theta [IAF − 6 ÷ IAF − 2 Hz], alpha [IAF − 2 ÷ IAF + 2 Hz], beta [IAF + 2 ÷ IAF + 16 Hz], and gamma [IAF + 16 ÷ IAF + 25 Hz]. The recording sessions were segmented into trials, corresponding to the ms of listening to the stimuli. The scalp EEG Power Spectral Density (PSD) was calculated in correspondence to the different conditions with a frequency resolution of 0.5 Hz. The normalisation of the trials was carried out by subtracting the open-eye brain activity recorded before the start of the experimental task.

The Workload Index (WI) was defined as the ratio between EEG PSD in Theta band over the central frontal area (Fz, F7, F8, F3, F4) and the EEG PSD in alpha band over the central parietal area (Pz, P7, P8, P3, P4) [56–58]. We use WI to evaluate its effectiveness in gauging the 'musical effort' recently defined [30] as 'the cognitive workload that the act of imagining music, playing it, or listening to it, imposes on the mind or brain of a professional pianist as well as other individuals with different musical expertise'.

To monitor the pleasantness was used the Approach Withdrawal Index (AW), synthesising frontal asymmetry in alpha band, defined as follows:

$$AW = GFP\alpha_{right} - GFP\alpha_{leftAW} = GFP\alpha_{right} - GFP\alpha_{left}$$

where the GFPα_right and GFPα_left stand for the GFP calculated among right (F4, F8) and left (F3, F7) electrodes, respectively, in the alpha (α) band.

In line with the pioneering frontal EEG asymmetry model of Davidson [59,60] and recent applications in clinical and non-clinical populations [61–64] positive values indicate relative greater right alpha frontal activity suggesting an approach tendency; vice versa, negative values would underline relative higher left alpha frontal activity, suggesting a withdrawal tendency.

2.6. Statistical analysis

Inferential statistics were applicated: both neurophysiological and behavioural data were compared between and within NH and UCI groups through analysis of variance (ANOVA). The dependent variables were represented respectively by WI, AW employed to estimate the neurophysiological activity and RT, ACC, DIFFICULTY, PLEASANTESS employed to capture behavioural and cognitive patterns; the independent factor were respectively, the SNR condition (Q, V, X) and the EMOTION condition (H, S). The statistical analysis was completed by performing the post-hoc Duncan's test at the 0.05 level of significance. A Pearson's correlation was performed to investigate the relations between behavioural and neurophysiological data.

3. Results

3.1. Behavioural results

RTs and ACC data are presented in Table 2.

Anova results in term of ACC, showed a statistically significant difference between SNR conditions [F (2,24) =4.925, $p = .016$, $\eta_p^2 = 0.291$]. The highest significant percentages of correct responses were measured for X compared to V ($p = .01$) and Q ($p = .01$) conditions. With respect to the group, overall NH performed statistically better than UCI [F (1,12) = 6.851, $p = .022$, $\eta_p^2 = 0.363$]. Furthermore, the Post hoc conducted after the evidence of statistical interaction between SNR condition and Group [F (2,24) = 4.333, $p = .024$, $\eta_p^2 = 0.265$] showed that UCI, compared to NH had a worse performance in condition

Table 2. Behavioural data.

		Behavioural Data (Mean ± s.d.)				
		Experimental conditions				
		Quiet (Q)	SNR + 5 (V)	SNR + 10 (X)	Emotion-Happy (H)	EmotionSad (S)
RTs	TOT	1712.875 ± 823.880	1873.232 ± 754.250	1845.661 ± 861.513	1914.643 ± 965.193	1706.536 ± 658.687
	NH	1539.482 ± 406.542	1630.125 ± 423.243	1487.071 ± 222.726	1482.357 ± 339.235	1622.095 ± 340.700
	UCI	1886.268 ± 111.422	2116.339 ± 994.051	2204.250 ± 1121.815	2346.929 ± 1211.342	1790.976 ± 898.518
ACC	TOT	0.758 ± 0.210	0.750 ± 0.264	0.857 ± 0.175	0.845 ± 0.226	0.732 ± 0.275
	NH	0.857 ± 0.168	0.928 ± 0.141	0.928 ± 0.141	0.988 ± 0.031	0.821 ± 0.265
	UCI	0.660 ± 0.213	0.571 ± 0.237	0.785 ± 0.187	0.702 ± 0.249	0.642 ± 0.275

V both when considering the same condition ($p = .004$) and when comparing with conditions Q ($p = .014$) and X ($p = .003$) respectively. Interestingly, no difference in accuracy in quiet ($p = .07$) was found.

Considering the differences in ACC *within* groups under different SNRs, NH did not differ while UCI showed better performances in X than in Q ($p = .028$) and V ($p < .001$) SNR levels.

Regarding RTs there was a statistically significant effect of the EMOTION condition on the group (NH/UCI) factor [F (1,12) =8.159, $p = .014$, $\eta_p^2 = 0.404$]: post hoc analysis revealed that UCI had longer RTs in the H condition than in S condition ($p = .007$).

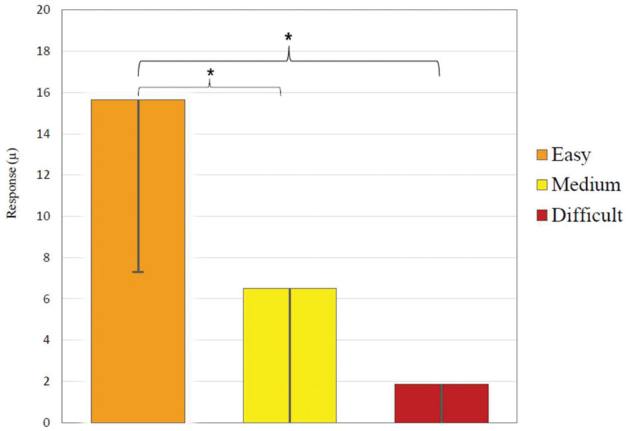

Figure 2. Representation of the cognitive evaluation of stimuli difficulty. Error bars stand for standard deviations. Significant differences between evaluations emerging from post hoc test are indicated (**$p \leq .01$; ***$p \leq .001$).

In terms of DIFFICULTY, there was a significant difference between the degree of perception [F (2,24) = 10.280, $p < .001$, $\eta_p^2 = 0.461$]; post hoc analysis showed that, overall, the task was rated significantly *easier* than *medium* ($p = .007$) and *difficult* ($p < .001$), regardless of groups and conditions (Figure 2).

In terms of PLEASANTNESS [F (2,24) =13.452, $p < .001$, $\eta_p^2 = 0.528$] post hoc analysis revealed that the task was globally considered more *pleasant* than *indifferent* ($p < .001$) and *unpleasant* ($p < .001$). Finally, Anova test showed a significant interaction between self-reported level of pleasantness and SNR condition with respect to group (NH/UCI) [F (4,48) =5.3241, $p = .001$, $\eta_p^2 = 0.307$]. The post hoc results are depicted in Figure 3.

3.2. Neurophysiological results

Concerning WI, there were no statistically significant differences between groups neither in the SNR nor in the EMOTION condition (all $p > .05$).

The AW results evidenced higher values for UCI compared to NH [F (1,12) =4.817, $p = .048$, $\eta_p^2 = 0.286$] and a statistically significant interaction between SNR condition and Groups [F (2, 24) =3.302, $p = .050$, $\eta_p^2 = 0.215$). Post hoc analysis (Figure 4) showed significantly lower values *within*

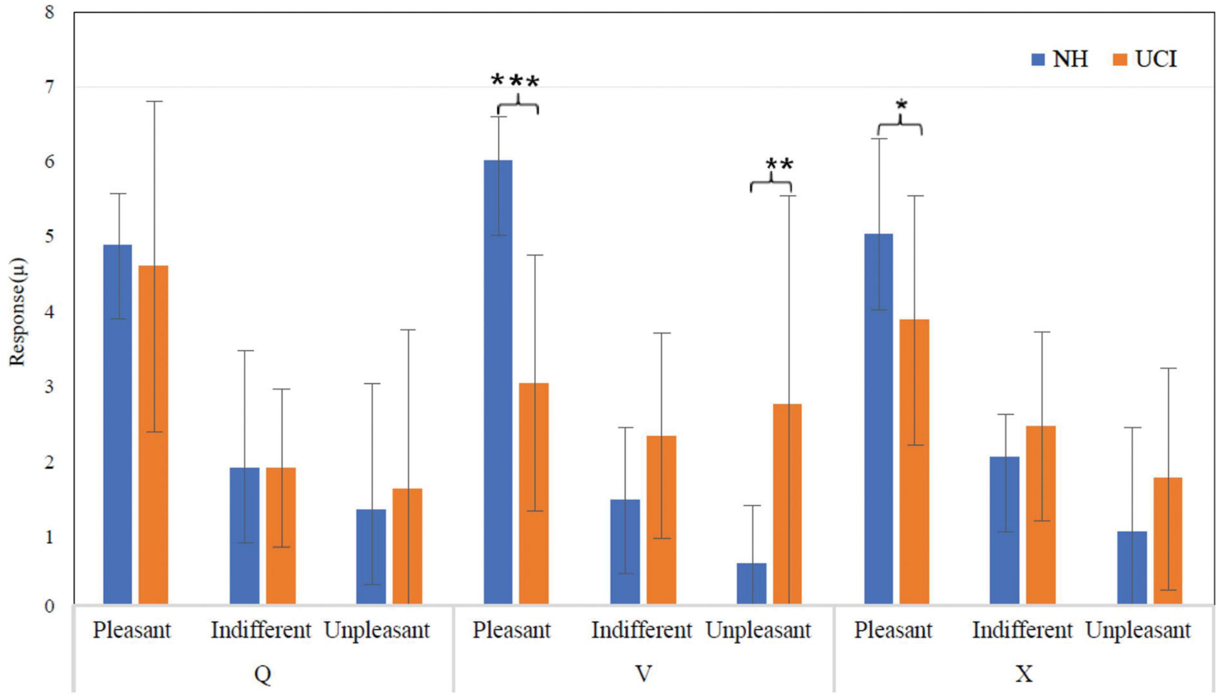

Figure 3. Cognitive evaluation of stimuli pleasantness in the 3 SNR conditions (Q,V,X). Error bars stand for standard deviations. Significance between and within Groups (NH; UCI) is highlighted (*$p \leq .05$; **$p \leq .01$; ***$p \leq .001$).

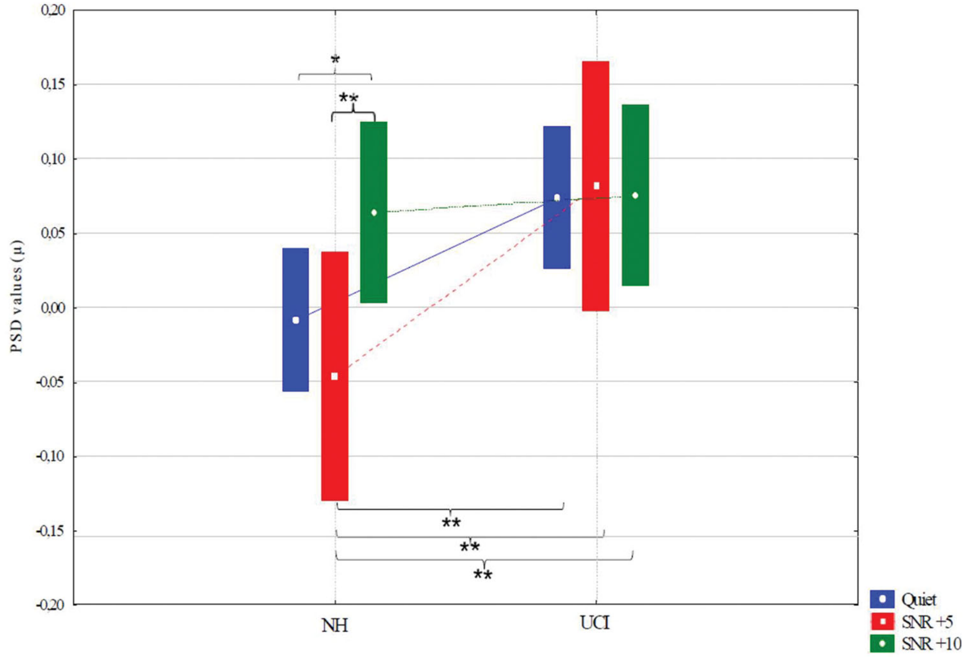

Figure 4. AW Index in noise conditions. Error bars stand for standard deviations. Statistically significant differences between/within groups (NH; UCI) are indicated ($*p \leq .05$; $**p \leq .01$).

NH in V than in X condition ($p = .003$) with higher values in the latter than in the Q condition ($p = .034$). Contrarily, no difference for AW emerged *within* UCI group ($p > .05$), while occurred *between* UCI and NH: in the noisiest condition (V) NH had lower values than UCI with respect to Q, V and X ($p = .014$; $p = .012$; $p = .015$).

Correlation results, in the noisiest condition, provided evidence of a significant relationship between WI and happy stimuli (r = −0.82).

4. Discussion

In this pilot study, a neurophysiological approach to the estimation of the perceived 'musical effort' and 'musical pleasantness' during the listening of classical emotional music excerpts presented in different noise conditions, in UCI and NH adults, was presented using two EEG indeces: WI and AW respectively.

Although RTs globally increased with SNR level, the noise did not significantly affect RTs either in the total population or *between* and *within* groups. This result, apparently in contrast with literature showing that background noise is detrimental to tasks involving cognition, concentration and attention [65], is neurophysiologically confirmed by the trend observed in WI. In fact, the explorative analysis conducted to evaluate musical effort shows that noise did not have a significant impact on brain activity either in the total population or *between* and *within* groups. A possible explicative hypothesis could be that the task was not particularly tiring compared to other noise listening tasks, perhaps due to the previously demonstrated beneficial effect of music on cognition [66]. This suggestion is supported by the evidence that auditory stimulation overall was perceived as *easier* than '*medium*' or *difficult*, irrespective of conditions and groups. Therefore, the cognitive data in terms of perceived difficulty seems to support the validity of the WI as a measure of musical effort.

In terms of ACC, more correct answers occurred in the medium than in the noisier and quieter conditions ($p = .01$). Furthermore, while in agreement with other studies [35] NH did not show significant differences between SNRs, UCIs showed a significant improvement in accuracy in X compared to quiet and noisier conditions. This pattern seems to be in line with past studies that demonstrated that noise may improve simple vigilance tasks [67]. Further, these results seem to corroborate the counterintuitive stochastic resonance (SR) phenomenon whereby an optimal amount of noise may under certain circumstances be beneficial for cognitive performance [68]. Accordingly, SR might contribute to improve signal detection in the auditory system [69] as well as in CI processing [70].

Emotions had a significant impact on RTs: UCI showed longer RTs for happy musical extracts than for sad ones. This result can be discussed considering previous studies demonstrating CI users' difficulties in recognising vocal emotions [71,72]. In general, CIs more easily identified the sad, angry, and neutral target emotions than the happy and fearful targets [73]. In this perspective, the strong negative correlation between musical effort in terms of WI and happy excerpts could be a result of the patients' abandonment of the task. Presumably, the ability to identify emotional expressions in western music is partly due to the universal ability to recognise non-verbal patterns of emotional expressiveness [74] such as musical prosody, a complex, rule-governed form of auditory stimulation that can move listeners emotionally in a systematic way [75]. Differently from a hearing system with a natural cochlea, capable of transferring the temporal fine structure of the auditory stimulus, a bionic hearing system transmits mostly a temporal envelope [76], displaying a poorer processing of the fine temporal and spectral information [77]. In fact, although coarse spectro-temporal resolution provided by a CI is adequate for speech recognition [78] this is not sufficient for the acoustic cues in which musical prosody is instantiated (pitch, timbre, amplitude) [75,79,80]. Considering the aforementioned technical aspects of signal processing, the result of longer RTs recorded in UCIs for happy stimuli recognition, may lie precisely in the structural characteristics of the stimulus and in the ability of a monolateral stimulation in processing this aspect like a natural cochlea. A previous study [81] conducted on children with unilateral and bimodal CI with stimuli originating from the same database here used, showed that the presence of CI makes them inferior to NH peers in an emotional recognition task.

Our experimental design allowed us to compare both the component of difficulty due to background noise and that due to the emotional characteristics of music. Overall, performance results would seem to confirm the specific difficulty of processing musical emotion compared to background noise in this category of patient.

The analysis of pleasantness investigated through the AW suggests that the pattern of modulation of such cerebral index is in agreement with the frontal EEG asymmetry theory [60] only for NH. In fact, in the noisiest condition NH have lower values than in the quiet condition, thus an ecological tendency to withdraw from a perceived unpleasant stimulus, in accordance with previous studies [82].

Not surprisingly, the tendency to approach increases significantly in X compared to Q condition suggesting the ability of a natural auditory system to appreciate and enjoy music also in nonsilent environments (as it should be, thinking for example about the pleasure in listening to music in daily situations such us on the bus, during sport activities etc.). However, to more deeply understand this difference between Q and X conditions, further analysis of the qualitative characteristics of the musical excerpts (e.g. pitch, mode, tempo) and the sound spectrum of each stimulus would be required.

Anyway, these ecological patterns of brain activation are not observed in patients: UCIs perceive the stimulus but show a neurophysiological deficit in discriminating between conditions, findings that are directly in line with previous works [82,83]. Nevertheless, the lack of difference in AW in Q between groups, could indicate the healthy ability to neurophysiologically enjoy the pleasantness of the musical stimulus in silence also for UCI when music is simplified in terms of multi-instrumental perception.

Interestingly, the cognitive evaluation in terms of perceived pleasantness only partially reflect the brain patterns. In fact, while cognitive and neuroimaging results in quiet are mirrored, NHs rate noisy listening conditions more highly than UCIs. Additionally, for the noisiest condition, UCIs claim more unpleasantness than NHs. This cognitive evidence, opposite to AW values, suggests a difference between groups with respect to the immediate brain pleasantness response and the cognitive evaluation of the perceived stimulus. In fact, it could be assumed that the immediate (bottom-up) neurophysiological response of UCI is not able to identify differences among noise conditions, while later, through a cognitive appraisal (top-down), the patient is more able to differentiate among conditions and to express a subjective evaluation of pleasantness. Following this reasoning, it is logical that the NH has a greater ability to focus on the pleasant stimulus (music) even in noisy conditions, being able to attribute to the listening greater values of pleasantness than UCIs; while the bottom-up stimulus-mediated mechanism shows in the health auditory system, an atavistic immediate response of withdrawal from the most disturbing stimulus.

This interpretative channel could find confirmation by the absence of processing differences between groups when listening to music in quiet. The monoinstrumental musical stimulus (piano solo) perceived in quiet, indeed does not involve differences in either the neurophysiological or cognitive-behavioural patterns between natural and bionic auditory systems. This could lead to the conclusion that the listening pleasantness of the 'pure' musical excerpts is processed indifferently between NH and UCI both at the cortical and cognitive levels. These findings give experimental support to the observations that CI users can still be attracted to music and can enjoy music listening [17,84] even if in more complex listening conditions they cannot fully appreciate the music.

Finally, in addition to the foregoing, the absence of musical effort in noisy auditory conditions shows the CI effectiveness, although in a limited sample, in preserving brain resources that could be devoted to improving the enjoyment of music listening, because, as the results of this pilot study seem to suggest, even a bionic auditory system can be tuned for music.

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Smoke signals: A study of the neurophysiological reaction of smokers and non-smokers to smoking cues inserted into antismoking public service announcements

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A R T I C L E I N F O

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A B S T R A C T

Tobacco addiction is one of the biggest health emergencies in the world, Antismoking Public Service Announcements (PSAs) represent the main public tool against smoking; however, smoking-related cues (SCs) often included in PSAs can trigger ambiguous cerebral reactions that could impact the persuasiveness and efficacy of the antismoking message.

This study aimed to investigate the electroencephalographic (EEG) response in adult smokers and non-smokers during the exposure to SCs presented in antismoking PSAs video, in order to identify eventual neurophysiological features of SCs' 'boomerang effect' elicited in smokers.

EEG frontal Alpha asymmetry and frontal Theta were analyzed in 92 adults (30 no smokers, 31 low smokers, 31 high smokers) from EEG recorded during the vision of 3 antismoking PSAs, statistical analysis was conducted using ANOVA.

Main results showed a significant interaction between smoking cue condition (Pre and Post) and smoking habit (in particular for female heavy smokers) for the frontal Alpha asymmetry. Since the relative higher right frontal Alpha activity is associated with approach towards a stimulus, it is suggested that the relative left frontal Alpha increase in response to SCs might reflect an appetitive approach in response to it. In the light of the Incentive Sensitization Theory, this pattern can be interpreted as a neurophysiological signal in response to SCs that could undermine the message's effectiveness contributing to the maintenance of the addiction.

1. Introduction

Tobacco smoke is the second leading cause of death in the world and the first reason of preventable death killing eight million people each year, through heart disease, lung cancer and other illnesses, plus passive smoking (WHO report, 2019[2]).

Numerous public measures have been set up to fight this problem (National Cancer Institute, 2018[3]) and Public Service Announcements (PSAs) represent one of the most effective strategies to influence citizen behavioral changes (Hornik, 2002; Davis et al., 2008; Lee et al., 2011).

DSM 5th edition includes tobacco addiction in "Substance-related disorders" category, recognizing nicotine's ability to develop addiction in humans (American Psychiatric Association, 2013; Benowitz, 2010). Scientific evidence shows that nicotine acts on the meso-limbic

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[2] WHO Tobacco global Report: https://www.who.int/tobacco/global_report/en/.

[3] https://www.cancer.gov.

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dopamine system (Nestler, 2001, 2005; Wise, 2004; Koob and Le Moal, 2001; Koob and Wolkow, 2010; Hyman et al., 2006), that projects from the Ventral Tegmental Area (VTA) to the Nucleus Accubens (NAc) and has been implicated in the rewarding effects of drugs abuse (Wise and Bozarth, 1987; Koob, 1992; Wise, 1996; McBride et al., 1999; Pierce and Kumaresan, 2006).

Nicotinic receptors in the meso-limbic system therefore constitutes the fundamental neurobiological substratum in the mediation of the gratifying effects of nicotine and of the development of tobacco addiction (Pidoplichko et al., 2004; Balfour, 2004).

Neuroimaging investigations provide tangible confirmations of how smoking induces dopamine release in the NAc (Brody et al., 2002; Sharma and Brody, 2009) and so how nicotine allows the establishment of pathological dependency behaviors (Carter and Tiffany, 1999). However, numerous factors including genetic baggage (Agrawall et al., 2012; Bierut et al., 2007), gene-environment interactions in development (Tully et al., 2010; Kendler et al., 2008) personality traits (Terraciano and Costa, 2004; Zuckermann et al., 1990), metacognitions about smoking (Nikčević and Spada, 2010) psychosocial factors (Chassin et al., 2000; White et al., 2002), age (DeBry and Tiffany, 2008) and mental illness (Prochaska et al., 2017) contribute or co-occur with addiction.

Nicotine is essential in addiction development, but only after extensive self-administration experience, while nicotine-associated cues can powerfully affect self-administration behaviors. Thus, although the dependent smoker may smoke to quell or achieve internal states (e.g., to reduce negative moods), external cues can powerfully influence self-administration behaviors (Baker et al., 2004). In fact, non-nicotinic factors play a fundamental role in addiction (Caggiula et al., 2001) as confirmed by the non-univocal results found in the therapeutic strategies directed to the action of nicotine - Nicotine Replacement Therapy (NRT) (Silagy et al., 2004).

Indeed a smoker is not a mere nicotine's self-administrator: many stimuli are associated with smoking behavior (Benowitz, 2009). Smoking desire is triggered by stimuli associated to nicotine's intake, such as people, environments and mood status (Tiffany & Tiffany and Drobes, 1990; Michalowski and Erblich, 2014; Conklin et al., 2015). So, a “Smoking Cue (SC)” is defined as a visual cue eliciting smoking urges in adult smokers and that represents at least one of the following: (a) smoking-related materials (i.e., cigarettes, ashtrays), (b) holding and handling of a cigarette without smoking it, and (c) actual smoking of a cigarette (Kang et al., 2009; Hutchinson et al., 1999; Tiffany et al., 2000; Waters et al., 2004).

SCs' role in relapse contrast in smokers wishing to quit is well known (Ikard et al., 1969; Droungas et al., 1995; Shiffman et al., 2013; Shiffman, 1982). The development of automatic associative processes with repeated tobacco-use could be as powerful as the direct effects of smoking (Brunzell and Picciotto, 2009). The basis of nicotine addiction is a combination of positive reinforcements, including enhancement of mood and avoidance of withdrawal symptoms, and conditioning has a crucial role in the development of tobacco addiction (Benowitz, 2009).

Classical pavlovian conditioning paradigm (Pavlov, 1927) offers the theoretical framework for understanding the learning processes developed in nicotine addiction (Bevins and Palmatier, 2004; Bevins and Murray, 2011; USDHHS Report, 2010).

Conditioned stimuli (CSs) to cigarettes smoke act as motivational magnets following repeated association with unconditioned stimuli (UCSs) (Berridge et al., 2009).

According to Robinson and Berridge's “Incentive Sensitization Theory” (IST), repeated exposure to potentially addictive substances such as nicotine can induce brain changes to circuits that mediate the psychological processes involved in motivated behavior: the attribution of incentive salience to CS (Robinson and Berridge, 2008). Addicted behaviors are due to progressive and persistent neuroadaptations caused by the repeated use of substances: changes in nervous system manifest both neurochemically and behaviorally through the phenomenon of “sensitization”, defined as hypersensitivity to incentive motivational effects of drugs and their associated stimuli (Robinson and Berridge, 2003).

Over time, the mesolimbic dopamine system's sensitization transforms drug-related cues in motivational magnets (Berridge et al., 2009; Robinson and Berridge, 2013). Repeated pavlovian CS-UCS association increases the predictive value of the CS relative to the UCS, a process that can be described in terms of correlation and computational models (Anselme et al., 2013).

This theoretical framework could explain how smokers, despite the awareness of nicotine's danger, continue smoking compulsively often not feeling any pleasure and failing to explain the motivations of their behavior (Baker et al., 2004). Moreover, since the neuroadaptation induced by the substance is often permanent, it is hypothesized that is precisely the presence of the sensitization connected to these brain modifications of reward's circuit that makes smokers particularly sensitive to drug-related cues, causing relapse with evident clinical implications (Robinson and Berridge, 2000).

IST's pillars are reflected in experimental neurophysiological designs based on the reactivity of smokers to SCs. Winkler et al. (2011) clearly support how a neutral stimulus can elicit preparatory physiological responses if associated with smoking. Carter and Tiffany (1999) support the value of the experimental paradigm of cue reactivity by emphasizing how it induces physiological responses and craving in smokers and is therefore a valuable tool for investigating smoke addiction. Further confirmations are obtained from studies carried out through electroencephalography (EEG). Zinser et al. (1999) observe in smokers, a greater activation of the left hemisphere to the vision of SCs: results would therefore be congruent with the IST, representing the demonstration that approach motivation connected to the increased activation of the left frontal cortex (Davidson, 2004) is sensitive to stimuli associated with the rewarding substance. Studies in humans, show how levels of nicotine addiction influence electrophysiological activity (Haarer and Polich, 2000) and fMRI studies highlight how tobacco advertisement elicits cigarette craving and different neuronal activity in response to SCs primarily in smokers, indicating that they might be particularly responsive towards external SCs (Vollstädt-Klein et al., 2011). Moreover, literature illustrates how nicotine administration increases EEG activity shifting from low-frequency to high-frequencies activity (Teneggi et al., 2004; Knott, 2001; Domino, 2003) and how smoking deprivation is associated with shifts regarding the balance of Alpha activity between the left versus the right frontal hemisphere (Zinser et al., 1999).

The aim of the study is to assess, through EEG patterns, the possible ineffectiveness of media intervention, (for a comparative analysis of antismoking PSA see Wakefield et al., 2003) depending on target population's smoking habit.

In accord to Incentive Sensitization Theory basis here it is therefore hypothesized that heavy smokers would present a significantly different electrophysiological profile in comparison to light and non-smokers during antismoking PSA's vision depending on the appearance of SCs, and hence of the possible “boomerang effect” produced by them (Harris et al., 2014).

It is important to state that to our best knowledge, any previous EEG study investigated the reactivity to SCs in antismoking PSAs in the light of the IST.

2. Method

2.1. Participants

92 adult volunteers were recruited (46 M, 46F; average age = 34.70 ± 0.08 years old) subdivided in 31 Heavy Smokers (HS - 16 M, 15F), 31 Light Smokers (LS - 13 M, 18F) and 30 Non-Smokers (NS - 17 M, 13F). Participants smoking >5 cigarettes per day were classified as HS (Shiffman and Paty, 2006; Shiffman, 2009; Schane et al., 2010; Husten, 2009; Bjartveit and Tverdal, 2005); participants who did not smoke any cigarette were classified as NS. Subjects received detailed information

on the study and signed an informed consent. The experiment was performed in accord to the principles outlined in the Declaration of Helsinki of 1975, as revised in 2000, and it was approved by the Sapienza University of Rome ethical committee in charge for the study.

2.2. Stimuli

Participants watched 3 antismoking PSAs videos without sound displayed in a randomized order. Stimuli were presented on a 19″ flat screen with a distance from the subject varying from 50 to 60 cm. The PSAs were selected from an archive of 11 PSAs classified through an analysis conducted on antismoking PSA's literature diffused worldwide over the last twenty years. For the definition of the archive, the Key Performance Indicators (KPIs) stated by Varcoe (2004) and by Coffman (2002) were used. The 3 PSAs used here have been evaluated on the basis of the presence of smoking cues and of the prevailing emotion in the communication style. Then PSAs were divided into two parts (Pre-Cue and Post-Cue) based on SC' appearance (see Appendix for video's frames). The selected PSAs were:

i “The Breath Holder” (Havas Worldwide Helsinki advertising agency for Cancer Society of Finland, 2014): centered on happiness;
ii “Baby Love” (Havas Worldwide Helsinki advertising agency for Cancer Society of Finland, 2013): focused on fear (evoked by gruesome images and cold colors);
iii “Tobacco is Wacko” (Lorillard tobacco company, 1999): centered on disgust.

2.3. Experimental set-up

During the protocol execution, participants were sitting on a comfortable chair in front of a computer screen and they were not instructed with any particular task, just to be relaxed and to restrict head and body movements as much as possible to reduce the amount of extraneous noise in the EEG recordings.

Before starting, to remove the excess lipid layer from their skin, a solvent solution was passed on the forehead and on the ear lobes.

2.3.1. Electrophysiological signal detection

The EEG activity was recorded by means of a portable 19-channel system (BEmicro, EBNeuro, Italy) using a ten electrodes based EEG frontal band (Fpz, Fp1, Fp2, AFz, AF3, AF4, AF5, AF6, AF7, AF8) as indicated by the 10–20 International System (Jasper, 1958). Two reference auricular electrodes were respectively placed, one on the left auricular lobe (reference) and one on the right (ground). To facilitate signal's acquisition each electrode was filled with an electroconductive gel ensuring a constant electrode-skin adhesion. The EEG signal was acquired at a sampling rate of 256 Hz keeping the individual impedances below 10kΩ. After the preparation, asking the participants to fix a white cross on a black background on the screen for 1 min, the physiological variation at rest was acquired; furthermore as a baseline at the beginning and at the end of the stimuli sequence a low emotional impact documentary was used (Cartocci et al., 2016, 2017, 2019). At the end of the registration the equipment was removed and the residues left by the electroconductive gel were cleaned from the skin of each individual.

2.4. Data processing

In the pre-processing session it was possible to identify and remove the components of the track due to muscular artifacts, eye movements, blinks and recording-related noise. For this purpose a notch filter (50 Hz) was applied in order to eliminate the main current interference, than the gathered signal has been digitally band-pass filtered by a 5th order.

Butterworth filter ([2÷30] Hz), in order to reject the continuous component as well as high- frequencies interferences, such as muscular artifacts (Nuwer et al., 1998; Tsuchida et al., 2016). The Independent Component Analysis (ICA) procedure (Delorme and Makeig, 2004), allowed to detect and remove the independent components due to eye movement, blinks; finally the signal was recomposed. For each of the 92 participants, Individual Alpha Frequency (IAF) (Goljahani et al., 2012) was estimated on EEG at rest acquired at the beginning of the experimental task. Each band of interest has been defined as IAF ± X, expressed in Hz and being X an integer in the frequency domain (Klimesch, 1999), specifically:

- Alpha band = (IAF-3; IAF + 2).
- Theta band = (IAF-6; IAF-3).

For each participant, the individual calculation of the Global Field Power (GFP - Lehmann and Michel, 1990) Eq. (1) was calculated for each cortical area of interest.

$$\mathrm{GFP}(t)_a = \frac{1}{N}\sum_{1}^{N}\left(x_{i,a}(t)\right)^2 \tag{1}$$

The obtained Global Field Power (GFP) values, were normalized in *Z*-scores, referring to the baseline EEG activity.

2.5. Neurometric indices

Based on the acquired and processed EEG signal, appropriate evaluations were made to identify the variables to be considered in order to evaluate the brain's responses to the stimulations necessary for the research.

2.5.1. Alpha band: approach-withdrawal index (AW)

Extensive literature has shown that the frontal Alpha asymmetry between the activity of the two hemispheres implies a different motivational tendency towards a stimulus (e.g. Davidson et al., 1990; Davidson, 1992; Gray, 1977, 1978, 1990). In particular, two different neural systems mediate the approach and withdrawal motivation located in the left and right hemisphere respectively (Coan and Allen, 2004; Davidson and Irwin, 1999). Such studies confirm the prefrontal cortex (PFC) role in the implementation of positive (left PFC) and negative (right PFC) motivation (Davidson et al., 1990; Davidson and Irwin, 1999; Coan and Allen, 2004).

Several studies attribute to PFC a fundamental role in Approach/Withdrawal tendency in reaction to a wide range of stimuli (Di Flumeri et al., 2016; Maglione et al., 2015; Borghini et al., 2015; Schmidt and Hamslmayr, 2009; Balconi et al., 2009; Cartocci et al., 2018, Cartocci et al., 2019; Cherubino et al., 2019). The Approach-Withdrawal Index (AW) Eq. (2), derived from Davidson et al.'s (1990) studies was defined as follows:

$$AW = GFP\alpha\_right - GFP\alpha\_left \tag{2}$$

GFPα_right calculated over the electrodes Fp2, AF4, AF6, AF8, and GFPα_left over the electrodes Fp1, AF3, AF5, AF7, (Vecchiato et al., 2014). Each value obtained for the considered variables was standardized (*Z*-Score) using the baseline's average and standard deviation. Positive AW values indicate an approach tendency towards the stimulus, while negative AW values indicate negative motivation in response to a stimulus.

For each participant AW values were calculated for each second of acquisition during the stimulation; subsequently, averages of the AW Index were calculated in correspondence of the seconds related to the Pre-Cue and the Post-Cue phase.

2.5.2. Theta band

Frontal activity in Theta band has been considered to evaluate mental stimulus information's processing (Wascher et al., 2014; Wisniewski et al., 2015). A change in frontal Theta band activity is directly proportional to difficult' s level encountered processing a stimulus in different contexts (Berka et al., 2007; Cartocci et al., 2015; Wisniewski et al., 2015; Gevins and Smith, 2003; Borghini et al., 2017). Additionally, it has been already applied to antismoking PSAs study (Cartocci et al., 2016, 2017, 2019; Modica et al., 2017, 2018). Moreover recent studies show that low-theta EEG coherence in smokers' brains might be a biomarker of SC reactivity and can predict addiction behavior (Bu et al., 2019). Here, to evaluate mental processing were considered all frontal electrodes for GFP's computation for each second of video stimulation, after data standardization in Z-Score according to the baseline's average and standard deviation. Average Theta values were calculated in the Pre and Post-Cue phase.

2.6. Data analysis

For the comparison among the three stimuli, ANOVA test was performed considering as dependent variables AW Index and frontal Theta values. As independent variables were considered: "Smoking Cue" with two levels (Pre and Post); "Smoking Habit'" with 3 levels (NS, HS, LS); "Gender" (M, F). STATISTICA software was used for statistics elaboration, the post hoc Duncan's Test was used to perform pairwise comparisons on significant interactions.

3. Results

3.1. Approach withdrawal index (AW)

AW Index related to "The Breath Holder" stimulus highlighted a statistically significant interaction Smoking Cue × Gender F (1,83) = 5689, p =,019 and Cue × Smoking Habit × Gender, F (2,85) = 4121, p =,019. In particular, post hoc analysis revealed significant differences in the AW values: HS Females showed higher AW values both with respect to the NS Females (p =,029) and to HS Males (p =,027). Moreover, HS Females showed higher AW levels in the Post-Cue in comparison to the Pre-Cue segment (p =,011) (Fig. 1).

3.2. Frontal theta activity

Frontal Theta activity results showed a statistically significant effect in the interaction Smoking habit × Gender × Cue F(2,85) = 3255; p=,041 for "The Breath Holder" stimulus and the post hoc analysis highlighted a lower increase reported by NS Females in comparison to NS males during Post-Cue phase (p=,022) (Fig. 2).

Additionally there was a significant effect of the variable Smoking Cue in "Baby Love" stimulus F(1,85) = 12,136; p=,007 where we observe an increase in frontal Theta activity in Post-Cue, (Fig. 3) and in the interaction Smoking Cue × Smoking Habit × Gender F(2,85) = 4137, p=,019 where post hoc Duncan's test show higher activation in Post-Cue phase for the HS Female (p<,001).

4. Discussion

In this study, HS, LS and NS groups' frontal EEG Alpha and Theta oscillations were analyzed in response to SC presented in antismoking PSAs and it has been found a major activation in both bands in the Post-Cue phase in the HS female group. This finding could help to understand the 'boomerang effect' elicited by some antismoking PSAs (Harris et al., 2014) in the smoking habit because of the presence of SC.

It is known that exposure to cigarette-related cues is thought to precipitate smoking relapse (Shiffman et al., 1996) and that self-reported measures cannot provide reliable information about the brain mechanisms activated by drug cues (Cui et al., 2013). From Robinson and Berridge study (2003) it is suggested that such relapse could be linked to a high level of incentive salience, and that a measure of approach motivation (relative higher activation of left frontal cortex), is sensitive to manipulation of smoking motivation (Zinser et al., 1999).

AW Index showed statistical significance in HS Females for the stimulus "The Breath Holder" (Fig. 1), suggesting that this group had a greater approach in the Post-Cue phase because more solicited by SC's presence in the video, that was a mother smoking a cigarette at home just when her kid was coming in. Hypothesis that seems to be supported by the approach tendency showed by HS Females whilst the avoidance motivation reported by NS Females. It could be hypothesized that HS Females are more motivationally stimulated (as indexed by the frontal left asymmetry evidenced by AW) by "The Breath Holder" because of their interest in the child who with sad expression holds his breath in response to the vision of the smoking mother, a sort of "instinct of maternal protection" (Hrdy, 1999; Panksepp, 2011). This finding is congruent with current models of addiction suggesting that drug cues acquire high level of motivational significance (Robinson and Berridge, 1993, 2003; Volkow et al., 2004), and in line with evidences by Zinser et al. (1999): smoking cues have the ability to activate approach's mechanism in the smokers identifiable in frontal left asymmetry. This pattern seems to be the reminiscence of dopaminergic activities in mesencephalic structures.

Obtained results also show a strong gender difference in the AW: values would confirm the greater effect of the SCs on the female gender compared to the males (Perkins et al., 2001), and therefore explaining the greater difficulty encountered by females in quitting (Perkins, 1996; Wetter et al., 1999).

Increased frontal Theta activity in the Post-Cue phase for NS Males in respect to NS Female (Fig. 2) could presuppose a gender difference allocation of cognitive resources in the vision of a characterized stimulus (the woman) (for gender differences in EEG see Güntekin and Başar, 2007; Corsi-Cabrera et al., 1993).

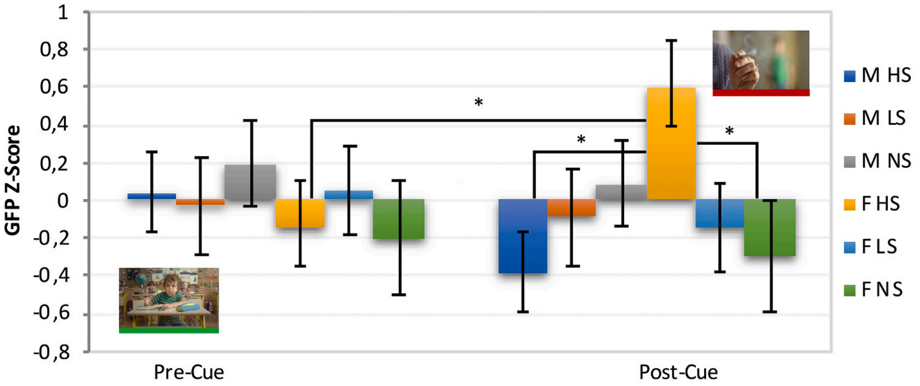

Fig. 1. Alpha Band Approach-Withdrawal Index (AW).
The graph represents the interaction among the variables Gender × Smoking Habit × Cue concerning the AW Index in "The Breath Holder" stimulus. Brackets stand for a statistical significance equal to $p \leq 0.05$. Error bars represent standard error.

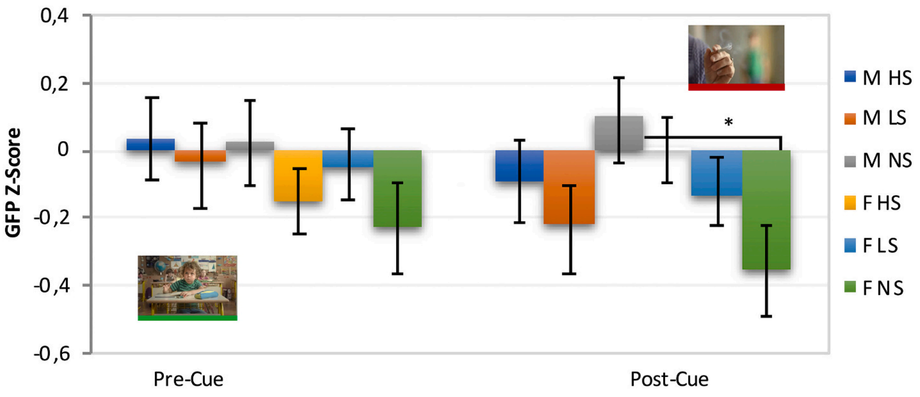

Fig. 2. Frontal Theta activity.
The graph represents the interaction among the variables Gender × Smoking Habit × Cue concerning the frontal Theta activity in “The Breath Older” stimulus. Brackets stand for a statistical significance equal to $p \leq 0.05$. Error bars represent standard error.

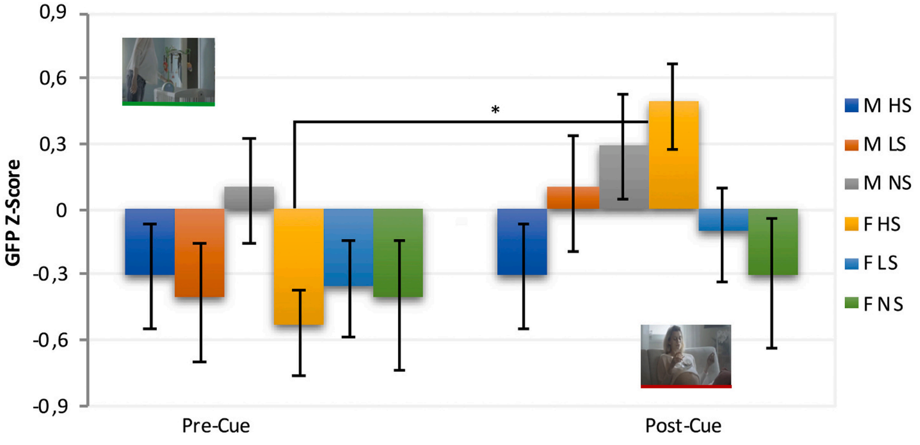

Fig. 3. Frontal Theta activity.
The graph represents the interaction among the variables Gender × Smoking Habit × Cue concerning the Frontal Theta activity in “Baby Love” stimulus. Brackets stand for a statistical significance equal to $p \leq 0.05$. Error bars represent standard error.

Furthermore, non-statistically significant activity in HS subjects in response to “The Breath Holder” video could be due to the craving elicited by SCs, that was already associated with a reduced Theta activation in smokers (Little et al., 2009). On the other hand, increased frontal Theta activity in HS Females in the Post-Cue phase during “Baby Love” stimulation (Fig. 3) could suggest a greater awareness of women's cognitive processing, in comparison to men, due to the strong message transmitted by the PSA, rather than a “wanting” activation induced by SC. Finally, SCs presence seems to increase the same activation framework (Theta synchronization) found during abstinence periods (Pickworth et al., 1989; Knott et al., 2008).

5. Conclusions

Results show that SC presence in antismoking PSAs have reliable effects on EEG frontal activation. Specifically, AW Index data confirm the initial hypothesis: according to IST, SCs inserted in PSAs built on a communicative style based on happiness, can elicit in HS (especially if females) different neurophysiological responses compared to a NS population. In particular, this could be interpreted as an activation signal related to smoking, possibly contributing to the maintenance of the addiction rather than its resolution.

SCs inclusion in PSAs must be evaluated with care and sensitivity to avoid their transformation into powerful motivational magnets able to convey, against a certain target, a message opposite to that desired. The “smoke signals” emerging from human neurophysiological substrates can therefore contribute to understanding and directing social intervention.

The present study is not without limitations. The first issue is the tobacco dependence assessment of the participant through standard instrument like Fagerström Test for Nicotine Dependence (FTND) (Fagerström, 1978) and the evaluation of craving smoking urge before and after video's stimulation (for example with Questionnaire on Smoking Urges (QSU) by Tiffany and Drobes, 1991).

Secondary, for better focus on the neurophysiological SC's impact during PSA vision, an additional protocol could be carried out modifying the same stimuli by replacing the presence of SCs with other neutral elements. In so doing, it would be possible undertake a comparison between Pre and Post smoking cue phase and Pre and Post neutral element phase in order to avoid interpretative bias connected to the perception of the stimulus.

Therefore the presents results constitute a premise for new research perspectives.

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Authors' contributions

BMSI and GC wrote the manuscript; BMSI, GC and EM performed the statistical analysis; BMSI and EM performed the data analysis; BMSI, GC, EM, DR and ACML performed the data recording; PC, LT, ACML and FB edited the manuscript; FB, supervised the project.

Declaration of competing interest

The authors declare no conflict of interest.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.ijpsycho.2021.06.010.

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OTOLOGY

The influence of auditory selective attention on linguistic outcomes in deaf and hard of hearing children with cochlear implants

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Abstract

Purpose Auditory selective attention (ASA) is crucial to focus on significant auditory stimuli without being distracted by irrelevant auditory signals and plays an important role in language development. The present study aimed to investigate the unique contribution of ASA to the linguistic levels achieved by a group of cochlear implanted (CI) children.

Methods Thirty-four CI children with a median age of 10.05 years were tested using both the "Batteria per la Valutazione dell'Attenzione Uditiva e della Memoria di Lavoro Fonologica nell'età evolutiva-VAUM-ELF" to assess their ASA skills, and two Italian standardized tests to measure lexical and morphosyntactic skills. A regression analysis, including demographic and audiological variables, was conducted to assess the unique contribution of ASA to language skills.

Results The percentages of CI children with adequate ASA performances ranged from 50 to 29.4%. Bilateral CI children performed better than their monolateral peers. ASA skills contributed significantly to linguistic skills, accounting alone for the 25% of the observed variance.

Conclusions The present findings are clinically relevant as they highlight the importance to assess ASA skills as early as possible, reflecting their important role in language development. Using simple clinical tools, ASA skills could be studied at early developmental stages. This may provide additional information to outcomes from traditional auditory tests and may allow us to implement specific training programs that could positively contribute to the development of neural mechanisms of ASA and, consequently, induce improvements in language skills.

Keywords Cochlear implants · Child · Auditory selective attention · Language skills

Introduction

Selective attention represents a fundamental cognitive capacity, that allows the brain to process targeted aspects of the environment, whilst simultaneously suppressing unwanted or distracting aspects [1]. It is critical for regulating external sensory inputs that occur within and across different sensory modalities, such as vision and somatosensory processing [2].

When this ability is referred to acoustic information, it is named auditory selective attention (ASA) [3]. ASA is crucial for everyday life as we live in a noisy environment where background sounds and human voices continuously overlap, requiring us to focus on significant stimuli in a particular moment, to avoid dangers (e.g., an incoming car if we are walking along the road) or to communicate with people (e.g., when listening to our own mother who is telling us a

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story) without being continuously distracted by irrelevant auditory signals.

In typical development, ASA is associated with children's lexical skills, explaining alone from 9 to 12% of variance in vocabulary scores obtained from six to seven years old children. ASA can be considered as an independent mediator in comparison to well-established factors that are significantly influencing vocabulary development (e.g., the verbal short term memory) [4]. Moreover, children with specific language impairment seem to show deficits in sustained selective attention tasks presented in the auditory modality under the high attentional load conditions, while showing similar performance to their typical developing peers on visual tasks regardless of the attentional load [5].

ASA is also highly relevant to the school setting in which instruction and completion of assignments may occur in a noisy environment with competing speech streams [6]. In situations that simulate noisy classroom settings, only children with good ASA are protected against the effects of noise in tasks where creative idea generation is required, in terms of showing performances similar to that they obtain in silence [7].

ASA depends on the ability to enhance the representation of an auditory source of interest. For this purpose, the listeners have to analyse the acoustic scene and to form a perceptual auditory object, i.e., a perceptual entity distinguished from other perceptual entities representing a stream of potentially interfering sounds [8, 9]. From this stream of sounds in an auditory scene, representing a mixture of individual sounds with various acoustic characteristics, the listeners should "segregate" the sound of interest, convey their attentional focus on it and ignore the interferers, whilst maintaining the cognitive flexibility to switch attention towards new auditory targets required by the context [9]. The entire process is complex for both adults and children. In adults, auditory object segregation and auditory attention are intertwined: the listeners need to have the ability to segregate the individual auditory stimuli that compose the complex auditory scene and to catch from it a potentially interesting sound, whilst the segregation process is biased by listeners' desire of attention [9]. During human development, instead, segregation of concurrent stream of sounds and auditory objects formation are the primary skills that allow infants to organize the auditory input around them, "thus enabling the development of cognitive abilities such as selective attention, speech perception (distinguishing speech from nonspeech sounds and separating concurrent streams of speech from each other), social skills, and memory (by distinguishing and, subsequently, correctly representing objects)" [10].

Both the segregation of concurrent stream of sounds and the appropriate formation of an auditory object depend on proper characteristics of the auditory signals, such as intensity, temporal/spectral structure, the onset/offset time, spatial cues and timbre features [11], as well as on subjective skills in processing binaural auditory information, such as summation, squelch, and head shadow effects [12]. Consequently, deaf and hard of hearing (DHH) subjects may show limited ability in auditory object formation due to the type, severity, and symmetry of the hearing deficit. This fact may variously affect their ability to detect acoustic signals and to perform a fine analysis of their temporal and spectral cues [13]. In turn, this may make it harder for them to perceptually segregate single components of the auditory scene [14]. The distorted formation of the auditory object negatively impacts the comparison and the differentiation of objects and consequently reduces the ability to suppress irrelevant ones [8].

In quiet environments, modern hearing devices can effectively overcome perceptual limitations resulted from degradation of processing in the peripheral auditory system [15]. In noise, instead, DHH subjects, face more challenging contexts that require an increased cognitive load to fill the perceptive gaps for processing acoustic information. The cognitive load is relative both to the use of top-down strategies to select the correct auditory object and to fill in continuously the gaps left by inaudible parts in the acoustic streams of information [8].

Studies on DHH adult populations show that subjects using hearing aids or cochlear implants (CIs) experience such difficulties [16, 17]. In particular, adult DHH CI users need to base the analysis of the auditory scene on the degree of perceptual differences between the stream, owing the limited spectral and temporal resolution of CI processing [15]. Limited signal processing negatively affects their ability to benefit from bilateral CI cues and acoustic segregation as well [18]. This leads to the formation of a less robust auditory object and may explain the difficulties that CI users still face in understanding speech in more challenging listening environments with multiple speakers, background noise and reverberation [19].

Limited dichotic auditory attention performance has been reported in bilaterally implanted DHH children [20, 21], with performance comparable to that of adult DHH CI users [21]. In fact, DHH children with bilateral CIs show a limited amount of unmasking when performing the dichotic test, characterised by the ability to ignore an interferer when presented to the ear opposite to the target and by binaural unmasking when the interferer is presented to both ears [21]. According to Misurelli et al. [21], these limitations might be caused by the poor peripheral encoding of speech signals that affect synchronous fusion of auditory images and central representation of the interferer.

No study up to now has investigated the influence of these limited ASA skills on language development in congenitally DHH CI children. In this context, the aim of the present study was to investigate and determine the unique contribution of ASA on the linguistic levels reached by a group of congenitally and profoundly deaf children of school age. Here, the effects of ASA were studied in respect to other personal and audiological variables that were traditionally considered to influence postoperative CI outcomes, e.g., nonverbal intelligence quotient (NVQI), age at diagnosis/implantation, family economic income (EI), maternal level of education (MLE) and auditory skills.

Differences in ASA may represent a further factor that may explain the high variability in linguistic outcomes after cochlear implantation in DHH children and this aspect needs to be investigated.

Materials and methods

The present research was a cross-sectional study, based on the rules of the STrengthening the Reporting of OBservational studies in Epidemiology (STROBE) statement (https://www.strobe-statement.org/, last accessed 10/02/2022). The protocol was approved by the local ethics committees of the two Cochlear Implant Centers that cooperated for the study's implementation and realization (Policlinico Umberto I Hospital, Roma; Guglielmo da Saliceto Hospital, Piacenza). The recruited families gave written informed consent for their own child's assessment before commencing any study-related procedure.

Table 1 Demographic and clinical characteristics of the study population ($n = 34$)

Variables	Median	Range
Age at assessment (years)	10.05	8–13.5
Age at diagnosis (months)	11.50	2–60
Age at CI (months)	18.5	7–66
Pre-CI PTA (dB HL)	101	93–110
Post-CI PTA (dB HL)	32	15–35
CPM normal score (percentile)	80	37–97
		n (%)
Gender		
Male		13 (38.2)
Female		21 (61.8)
Listening mode		
Monoaural CI		19 (55.9)
Bilateral CI		15 (44.1)
EI level		
Low		6 (17.7)
Middle–low		5 (14.7)
Middle		11 (32.3)
Middle–high		4 (11.8)
High		8 (23.5)
MLE		
Low (8 years)		5 (14.7)
Middle (13 years)		15 (44.1)
High (18 years)		14 (41.2)

Participants

Thirty-four DHH CI children (21 females, 13 males) with a median age of 10.05 years (range 8–13.5 years) were included. They came from different Regions of Italy (North, Centre, and South) and were enrolled in two Cochlear Implant Centers. Table 1 showed their main demographic and clinical characteristics.

All children had bilateral congenital profound sensorineural hearing loss, with a median preoperative pure tone average (PTA) of 101.5 dB HL (range 93–110 dB HL). Etiology of their hearing loss was as follows: unknown ($n = 15$), Connexin 26 mutation ($n = 17$), ototoxicity ($n = 1$) cytomegalovirus infection ($n = 1$). The median chronological age at diagnosis was 11.5 months (range 2–60 months), while the median age at implantation was 18.5 months (range 7–66 months). The median duration of device use at the time of assessment was 8.7 years (range 6–12.8 years).

Seventeen recipients were implanted with Cochlear devices that were fitted with ACE strategy, whilst 17 participants received Advanced Bionics devices and used Hi-Res 120 strategy. Eighteen children were bilateral CI recipients (9 simultaneous versus 9 sequential implantation), while 16 were monolateral CI users.

All CI recipients had normal cochlear conformation, with full insertion of electrode array. The absence of Central Auditory Processing Disorders and normal NVQI were verified using the Raven's Colored Progressive Matrices-CMP [22] for children up to 11 years of age and the Raven's Progressive Matrices-RPM [20] for children between 12 and 13 years of age. The sample's median normalized score at CPM and CMP was 80 (range 37–97).

Finally, all DHH CI children lived in monolingual native Italian-speaking environment, participated in oral rehabilitation programs, used auditory–verbal communication and were included in mainstream schools with a support teacher provided by the normal legislative procedure of Italian Ministry of Education.

Information concerning family economic income (EI) and maternal level of education (MLE) were gathered from their parents. EI was defined on the base of Italian economic family status indicator named as ISEE index (Indicatore della Situazione Economica Equivalente: Equivalent Economic Situation Index). The ISEE index based the allocation in the EI brackets computing the annual income, the real estate asset, the number of family members and the city of residence (https://www.inps.it/nuovoportaleinps/default.aspx?itemdir=50088#h3heading3). Based on this index, 5 EI brackets were defined: low, middle-low, middle, middle-high, and high. MLE was defined based on the years of formal education in three levels: low (8 years junior secondary school diploma), middle (13 years, senior secondary school diploma) and high (18 years, University degree). EI and MLE are detailed reported in Table 1.

Assessment

ASA assessment

ASA skills were assessed using the "Batteria per la Valutazione dell'Attenzione Uditiva e della Memoria di Lavoro Fonologica nell'età evolutiva-VAUM-ELF" [23]. Four dichotic listening tasks, that differed each other for the weight of the distraction's factor and for the level of cognitive workload, were used. For the distraction's factor, there was a condition with a medium linguistic interferer (the dichotic message was a piece of television News-N, less attractive for children) and another with a high linguistic interferer (the dichotic message was a Tale-T, more attractive for children). Regarding the cognitive workload, there were two consecutive conditions: an easier condition (ASA1) with a fixed target (the word "cane: dog)" and a more difficult condition (ASA2), where the target was a semantic category, specifically the "name of an animal". The four tasks that derived by the combination of linguistic interference and the cognitive workload conditions were: the fixed target CANE with the piece of television News as competitive message (ASA1-N); the fixed target CANE with the Tale as competitive message (ASA1-T); the target "name of an animal" with the piece of television News (ASA2-N) as competitive message; the target "name of an animal" with Tale as competitive message (ASA2-T). The difficulty of the task progressively increased from ASA1-N to ASA2-T. For every task, lists of bisyllabic words (8 target stimuli and 19 distractors) were used. The target stimuli were presented only once, while the distractors were repeated twice in random order, for a total of 46 words in each list. The duration of each test condition was 1 min and 15 s. The participant was requested to listen to the list and to raise the right hand when the target stimulus was presented, ignoring all the other words.

The tests were performed in a double-walled sound-treated booth, in sound field modality. The lists and the distractive messages were recorded and presented at the same level (65 dB SPL) through two loudspeakers positioned at 45° azimuth from the subject's head at a distance of 1 m- one loudspeaker for the distractive message and the other one for the target message. The lists containing the target were presented to the dominant ear: to the CI side in monolateral users and to the side with the best listening performance in bilateral or bimodal CI users.

The score was calculated on number of total errors (omissions or wrong target). Normal hearing children were shown to be able to perform ≤ 1 error at ≥ 8 years of age [23]. So, after this age, a score of 2 or more errors was indicative of selective attention difficulties.

Auditory skills assessment

Speech recognition in quiet was assessed by using standard phonetically balanced bisyllabic words for Italian pediatric population [24]. A 10-item test list was preceded by a practice list. Items were administered in a sound-proof room, via a loudspeaker placed ad 1 m distance from a table where the child was sitting next to a speech therapist. Speech stimuli were presented at 0° azimuth at 65 dB SPL, both in quiet and with speech noise fixed at + 5 Signal-to-Noise (*S*/*N*) ratio. The participant's score was calculated as the percentage of correctly repeated words.

The Categories of Auditory Performance-2 (CAP-2) was used to evaluate pediatric CI recipients' auditory outcomes in daily life. This tool has been a reliable measure of outcome, with a good inter-user reliability (correlation coefficient > 0.75) [25, 26]. The CAP-2 scale consisted of 9 categories in order of increasing difficulty:

0. No awareness of environmental sounds.
1. Awareness of environmental sounds.
2. Responds to speech sounds.
3. Identification of environmental sounds.
4. Discrimination of some speech sounds without lipreading.
5. Understanding of common phrases without lipreading.
6. Understanding of conversation without lipreading.
7. Use of phone with known listener.
8. Follows group conversation in a reverberant room or where there is some interfering noise, such as a classroom or restaurant.
9. Use of phone with unknown speaker in unpredictable context.

Language skills assessment

The DHH CI children were tested individually in a quiet room, by two female speech therapists. All children communicated verbally, so all tests were performed using spoken language.

TwoItalian Standardized Language tests were administered to assess lexical and morphosyntactic domains. Lexical comprehension was evaluated with the Italian version of Peabody Picture Vocabulary Test (PPVT), where normal standardized scores ranged from 85 to 115 [27]. Morphosyntactic comprehension was assessed with the Italian version of the Test for Reception of Grammar (TROG)-2 [28]. Based on its standard normative data, a score < 1 SD from the mean was considered as pathologic and this was indicated in the test's manual as the percentile ≤ 16°.

Italian version of PPVT [27] was an assessment tool that measured the receptive vocabulary in children. It consisted of 175 black and white stimulus items, displaying 4 pictures per page with increasing difficulty. The examiner said a word, and then the examinee responded by pointing out to the picture that s/he thought to correspond to the word presented by the examiner. The raw score was calculated by subtracting the number of errors from the highest number in the examinee's ceiling set. Test–retest reliability and internal consistency of the test were 0.93 and 0.94, respectively.

Italian version of TROG-2 [28] was a fully revised and re-standardized version of the widely used TROG, originally developed to investigate morpho-syntactic comprehension skills in children. The TROG-2 consisted of 20 blocks, each testing a specific grammatical construction, having an increasing order of difficulty. Each block contained four test items and the child needed to respond correctly to all of them to level up. Each test stimulus was presented in a four-picture, multiple-choice format with lexical and grammatical foils. For each item, the examiner read a sentence that referred to one of four drawings, and the participant's task was to point out to the drawing that corresponded to the meaning of the sentence. The score was calculated as total number of achieved blocks. Split-half reliability and internal consistency of the test were of 0.88 and 0.90, respectively.

Statistical analysis

Analyses were carried out using a PC version of Statistical Package for Social Sciences 25.0 (SPSS, Chicago, IL, USA). Sample characteristics were reported as average and standard deviation or median and minimum–maximum values, following the analysis of normality. DHH CI children's outcomes were compared with scale norms from the test batteries (obtained from nationally representative samples with typically developing, normally hearing children). The percentage of children performing within the normal range in the ASA tasks was reported. Wilcoxon's test was used to assess if there were statistically significant differences between ASA performances based on the degree of the task complexity (medium vs high linguistic interferers to competitive message and low vs high cognitive workload).

The Spearman's rank correlation coefficient was calculated to investigate the relations between the scores at the language and ASA tests, demographic characteristics (chronological age, NVQI), and audiological variables (age at diagnosis, age at implantation, duration of CI use, mono or bilateral listening, bisyllabic words recognition in quiet and in noise, CAP). Mann–Whitney and Kruskal–Wallis tests were performed to assess differences between gender, listening mode (mono and bilateral users) and mother level of education and economic income degree subgroups.

All variables with p values less than 0.05 in either direction were considered as significant and were afterwards used in a stepwise hierarchical multiple regression [29] to determine their contribution in predicting linguistic skills. The contribution was assessed in stages, allowing the systematic removal of different sources of information as well as the identification of the unique proportions of variance in the outcomes that could be identified with particular predictors. Variables entered on later stages are thus tested for their unique contribution after removing the contributions of earlier-entered variables.

Results

ASA skills

Median values and range of errors at the ASA tasks were reported in Table 2. The percentages of CI children with adequate performance were 50% in ASA1-N, 52.9% in ASA1-T, 38.2% in ASA2-N and 29.4% in ASA2-T. Both omission errors (61.4%) and wrong target errors (38.6%) were observed and their difference was statistically significant ($Z = -4.9$, $p < 0.001$, $\eta^2 = 0.706$; Cohen's $d = 3.101$).

No statistically significant differences were found in CI children's responses when comparing tasks with medium and high linguistic interferers (ASA1-T vs ASA1-N: $Z = -1.14$; $p = 0.25$; ASA2-T vs ASA2-N: $Z = -1.32$; $p = 0.18$).

Performances differed significantly for cognitive workload: the number of errors from the tasks with semantic category was significantly higher than those from the task with the fixed target (ASA1-T vs ASA2-T: $Z = -3.15$, $p = 0.002$, $\eta^2 = 0.292$; Cohen's $d = 1.284$; ASA1-N vs ASA2-N: $Z = -3.59$, $p < 0.001$, $\eta^2 = 0.379$; Cohen's $d = 1.563$).

Bilateral CI children performed better than monolateral CI children (U value SA1-N = 79, $p = 0.021$, $\eta^2 = 0.143$; Cohen's $d = 0.816$; U value SA1-T = 73, $p = 0.013$, $\eta^2 = 0.171$; Cohen's $d = 0.908$; U value SA2-N = 73.5, $p = 0.014$, $\eta^2 = 0.168$; Cohen's $d = 0.9$; U value SA2-T = 76.5, $p = 0.018$, $\eta^2 = 0.154$; Cohen's $d = 0.854$).

Listening and linguistic skills

Detailed scores for listening and linguistic skills were reported in Table 3.

Table 2 Auditory selective attention skills of the study group ($n = 34$)

Test	Median	Range
ASA1-N (n. errors)	1	0–7
ASA1-T (n. errors)	1.5	0–8
ASA2-N (n. errors)	2.5	0–8
ASA2-T (n. errors)	3	0–8

Table 3 Listening and language outcomes of the study population ($n = 34$)

	Median	Range
Word recognition in quiet (%)	100	60–100
Word recognition at + 5 *S*/*N* ratio (%)	80	10–100
CAP-2 (n. of category)	7	4–8
PPVT (normal standardized scores)	90	55–125
Trog-2 (percentile)	30°	1°–90°

Median bisyllabic words recognition percentages were 100% (range 60–100%) in quiet and 80% (range 10–100%) in the presence of speech noise at + 5 *S*/*N* ratio. Thirty children (88%) showed very high auditory performances ($\text{CAP} \geq 7$), reflecting the ability to communicate in more complex situations, such as noisy or reverberant environments or conversation at phone. The remaining four children (12%) showed a need to stay in a quiet setting; despite being poorer performers, they had anyway the ability to understand language without lipreading (CAP 4–6).

Standard Peabody median score was 90 (range 55–125) with 64.7% of children falling within the normal range for lexical comprehension. The median standard score at TROG-2 was 30° percentile (range 1°–90°) with 67.6% of children achieving normal scores for morpho-syntactic comprehension.

Relationships between ASA and language skills

ASA findings correlated significantly with all language tests (Table 4). The strength of their correlations with both lexical and morphosyntactic comprehensions were moderate. Likewise, all ASA subtests were strongly correlated to each other (all Rho scores > 0.8, $p < 0.001$) and the same was true for the test of language assessment (all Rho scores > 0.75, $p < 0.001$).

Owing these statistically significant correlations, a principal component analysis (PCA) was adopted to reduce the number of variables for further analysis [30]. The purpose of PCA was to derive weighted linear combinations of the individual measures that were strongly correlated, thus reducing redundancy in multiple regression analysis where the principal components were used as outcome variables. Components were more robust and representative of the study domain than any single test measure. Two new categories were identified: the linguistic component-LC (Peabody, TROG-2) and the ASA component-ASAC (ASA1-N/T, ASA2-N/T). Their Principal Components Loadings were shown in Table 5. In both analyses, PCA gave rise to one single component with a good efficiency, as indicated by the Kaiser–Meyer–Olkin (KMO) values (0.71 for LC and 0.743 for ASAC) and Bartlett p values (< 0.001 for both the components).

A new bivariate analysis was then performed to evaluate the correlations between the new components, and the results were still statistically significant (Rho = – 0.696, $p < 0.001$).

The unique contribution of ASA on language skills

A regression analysis was conducted to assess the unique contribution of ASA on language skills.

LC was used as the dependent variable and ASAC as the independent variable. For identification of other variables to be included in the regression analysis, significant factors in influencing LC were identified using Spearman correlation, Mann–Whitney, or Kruskal–Wallis tests depending on the nature of the variables.

Spearman's correlation test showed statistically significant effects of NVQI assessed by CPM as well as of age at diagnosis/implantation and listening skills (Table 6).

The Mann–Whitney test did not reveal any statistically significant differences in language performances neither because of gender ($U = 110.5$, $p = 0.347$), nor because of mono/bilateral listening mode ($U = 110$, $p = 0.241$). Mother's degree of education was analyzed depending on the achievement of a junior, high secondary school or university degree.

Table 4 Spearman's correlations between auditory selective attention tasks and language skills

	ASA1-N		ASA1-T		ASA2-N		ASA2-T	
	Rho	*p*	Rho	*p*	Rho	*p*	Rho	*p*
Peabody	– 0.51	**0.002**	– 0.49	**0.004**	– 0.46	**0.006**	– 0.49	**0.003**
TROG-2	– 0.51	**0.002**	– 0.55	**0.001**	– 0.40	**0.018**	– 0.47	**0.005**

Statistically significant values were set at $p < 0.05$ and are highlighted in bold

Table 5 Principal components loadings for auditory selective attention (ASAC) and linguistic (LC) components

Components	Loadings
Linguistic (LC)	
Peabody	0.94
TROG-2	0.94
Total variance explained 88.3%	
Auditory selective attention (ASAC)	
SA fixed target/tale	0.95
SA semantic target/tale	0.94
SA fixed target/news	0.93
SA semantic target/news	0.96
Total variance explained 89.9%	

The Kruskal–Wallis test showed that children from families with mothers of senior secondary school or university degree (13 and 18 years, respectively) had better linguistic skills than those with junior secondary school diploma ($H = 14.6$, $p = 0.001$ $\eta^2 = 0.389$; Cohen's $d = 1.596$).

ASAC and all these significant variables were added in the regression model as independent factors, using the stepwise method (Table 7). At the first step, only significantly effective demographic data, mother's level of education-MLE, and the children's characteristics (NVQI, age at diagnosis and at CI) were included. The only significant predictors were NVQI and age at diagnosis, which explained the 46% of variances. The earlier was the diagnosis and the higher was the intelligence quotient of the child, the better were the linguistic outcomes after cochlear implantation. At the second step, the speech perception in quiet/noise and CAP scores were included in the model. These variables together accounted for 8% of an additional variance in CI children's language competencies. The MLE and CAP scores were significant predictors: children with higher CAP scores and with mothers of a longer educational pathway, obtained the highest scores at language tests. Finally, at the third step of the model, the ASAC was added into the model in order to measure its unique contribution. This accounted for 25% of an additional variance and together with the other significant predictors–performance intelligence quotient, speech in noise and CAP–reached the 79% of the observed variances in linguistic skills of CI children.

Table 6 Spearman's correlations between linguistic component and demographic and audiological quantitative variables

	Linguistic component	
	Rho	*p*
Age at assessment	−0.20	0.25
NVQI	0.47	**0.005**
Age at diagnosis	−0.51	**0.002**
Age at CI	−0.42	**0.013**
Duration of CI use	0.073	0.68
Post-CI PTA	−0.08	0.96
CAP-2	0.59	**< 0.001**
Speech in quite	0.43	**0.01**
Speech noise	0.52	**0.001**

Statistically significant values were set at $p < 0.05$ and are highlighted in bold

Discussion

ASA is critical for learning and development during childhood. From the first day of birth, children receive spoken language in a complex listening environment where background noise is always present and may impair their ability to learn from the linguistic input, either by limiting the available resources for learning, or by making listening particularly straining [31]. Furthermore, background noise may distract children by leading to attentional shifts and information encoding failures, even with readily perceptible targets. Children struggle to learn words in background noise, particularly when the background noise consists of nontarget speech [32]. Noise has detrimental effects also on school achievements, since in school settings, the need to pay attention and to follow instructions or assignments that may occur in the presence of competing speech streams is essential [33]. The hearing children that are more skilled in processing the target stimuli while suppressing the information from other concurrent stimuli develop better verbal working memory, lexical and academic skills [4, 6, 7].

Table 7 Hierarchical regression analysis to establish the contribute of ASAC on LC

Variables	STEP 1	STEP 2	STEP 3
	β (*p*)	*β* (*p*)	*β* (*p*)
Maternal level education	0.24 (0.13)	**0.3 (0.04)**	0.19 (0.06)
NVQI	**0.36 (0.02)**	0.23 (0.11)	**0.37 (0.007)**
Age at diagnosis	**−0.43 (0.007)**	−0.17 (0.3)	0.03 (0.8)
Age at CI	−0.21 (0.51)	0.41 (0.8)	0.02 (0.9)
Speech in quite		−0.03 (0.8)	0.1 (0.36)
Speech in noise		0.3 (0.9)	**−0.59 (0.002)**
CAP-2		**0.55 (< 0.001)**	**0.67 (< 0.001)**
ASAC			**−0.67 (< 0.001)**
ΔR^2		0.10	0.20
R^2	0.46	0.54	0.79

Statistically significant values were set at $p < 0.05$ and are highlighted in bold

DHH postlingual adults and children with CIs show impaired ASA due to the CI's limited spectral resolution [17, 20, 21]. The present study confirms poor ASA skills in a DHH CI paediatric population with worst outcomes in monolateral CI users. Only 1 out of 2 DHH CI children achieves adequate performance for the less demanding ASA task (fixed target with medium linguistic interferer) and about 1 out of 3 performs within the normal range for the more complex task (changing target and high linguistic interferer), despite most of the children of the sample show good speech perception skills (CAP-2 ≥ 7). Most of the errors are represented by omission of the target and this fact allows us to speculate that the degree of perceptual discrimination between the target and the competitive message could be at the basis of the difficulty. Owing to the limited spectral resolution, DHH CI children may sometimes fail in forming the perceptual auditory object when the perceptual acoustic similarity between the target and the dichotic message makes the entire auditory target or it's portions imperceptible.

The present bilateral DHH CI children perform better than their monolateral peers on ASA tasks, similarly with the studies by Gordon et al. [34] and Misurelli et al. [21]. Having a bilateral CI helps DHH CI children to achieve their best performance in spatial hearing and in masking release, probably because of the availability of interaural level and timing cues, that are missing in monolateral listening condition [21, 34]. This in turn, despite high variability in the amount of release from masking between bilateral subjects [35], may condition language acquisition, since DHH children with bilateral CIs achieve significantly better vocabulary outcomes and significantly higher linguistic scores in comparison to monolateral users [36].

Regarding the contribution of ASA to linguistic skills, similarly with findings in children with typical development by Majerus et al. [4], ASA represents an independent contributor to oral language skills development in this sample of DHH CI children. ASA accounts for a 25% of variance to oral language outcomes in addition to the factors, such as cognitive level, maternal education level, early intervention, listening mode and speech perception skills that are traditionally considered when studying postoperative outcomes in paediatric CI users [37, 38]. As in hearing children, ASA seems to be implicated in language processing of DHH CI children. When children interact with other people and listen to spoken language, speech represents a complex acoustic signal, with rapidly changing stream of information having few objective boundaries. From this continuous stream of auditory input, then, children face the challenge of parsing word boundaries and extracting meaning. Furthermore, many speech sounds are discriminated mainly by subtle spectral or temporal differences on the order of tens of milliseconds and many morphemes have low perceptual salience in the context of continuous speech stream. Furthermore, the presence of environmental noise and distracting speech sounds complicate the perceptual task. Hence, it is reasonable that the ability to direct selectively the attention on a target message while ignoring and suppressing distracting information could help children to process language in a more facilitated way and this, in turn, could support them to develop better linguistic skills.

This research aims to be a first attempt in determining the impact of ASA on linguistic skills attained by DHH CI children but has several limitations due to the absence of prospective follow up, the small study sample size and the absence of tasks aiming to understand the cognitive and psychoacoustic processes that may explain the nature of its findings and the specific mechanisms of ASA in paediatric DHH CI population. For example, the development of the four components of attention, represented by arousal, orientation, allocation, and maintenance, have been studied in hearing populations and might be investigated in deaf children with CI as well [39]. Also, the use of purposely developed tasks, together with the event-related brain potential technique, may allow to examine the spectral and temporal dynamics of selective attention as observed in young typical developing hearing children by Astheimer and Sanders [40] or in hearing children with specific language impairment by Stevens et al. [41] and may give us new insights in how ASA works in DHH CI children.

Nevertheless, the present findings are clinically relevant as they highlight the importance to assess ASA skills as early as possible, reflecting their important role in language development. With simple clinical instruments, as in the present study, ASA skills could be studied at early developmental stages, even in children as young as 3 years [42]. This may provide us additional information to findings from traditional auditory tests and may allow us to gain insight into early implementation of specific training programs, that could induce improvements on standardized measures of language and contribute positively to the development of neural mechanisms of ASA [43]. In English-speaking children with specific language impairment, it has been observed that they may have difficulties with linguistic forms that are perceptually less salient, such as the past tense–ed inflection, possessive s or articles [44] and that improvements in the neural mechanisms of selective attention may facilitate perception and processing of these more vulnerable linguistics forms [43]. The early detection of ASA difficulties and the development of specific programs to train auditory attention in DHH CI children may represent a new challenge for clinicians in finding new tools for improving outcomes after cochlear implantation.

Finally, these findings suggest that even at the early postoperative phases, it is of the utmost importance to support DHH CI children with the most appropriate technology such as assistive listening devices [45, 46] or adaptive microphone systems [47] in order to improve S/N ratio in challenging listening environments, and to study the long-term effects on linguistic and academic skills.

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Article

Neuropsychological Functions and Audiological Findings in Elderly Cochlear Implant Users: The Role of Attention in Postoperative Performance

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Abstract: Objectives: The present study aimed to investigate in a group of elderly CI users working memory and attention, conventionally considered as predictors of better CI performance and to try to disentangle the effects of these cognitive domains on speech perception, finding potential markers of cognitive decline related to audiometric findings. Methods Thirty postlingually deafened CI users aged >60 underwent an audiological evaluation followed by a cognitive assessment of attention and verbal working memory. A correlation analysis was performed to evaluate the associations between cognitive variables while a simple regression investigated the relationships between cognitive and audiological variables. Comparative analysis was performed to compare variables on the basis of subjects' attention performance. Results: Attention was found to play a significant role in sound field and speech perception. Univariate analysis found a significant difference between poor and high attention performers, while regression analysis showed that attention significantly predicted recognition of words presented at Signal/Noise +10. Further, the high attention performers showed significantly higher scores than low attentional performers for all working memory tasks. Conclusion: Overall findings confirmed that a better cognitive performance may positively contribute to better speech perception outcomes, especially in complex listening situations. WM may play a crucial role in storage and processing of auditory-verbal stimuli and a robust attention may lead to better performance for speech perception in noise. Implementation of cognitive training in auditory rehabilitation of CI users should be investigated in order to improve cognitive and audiological performance in elderly CI users.

Keywords: cochlear implantation; cognition; older adults; speech perception; attention; working memory

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1. Introduction

In 2017, the Lancet Commission of Dementia Prevention, Intervention, and Care reported a new model of dementia risk [1,2] stating that hearing impairment may account for 8% of dementia cases [3]. Indeed, hearing loss (HL) is considered the biggest controllable risk factor for dementia, potentially reducing its risk by 9% [1]. This model originates from the growing evidence over the last decade highlighting the significant associations between presbycusis, deterioration of cognitive functions, and incident dementia [4]. The risk of incident dementia has been estimated to be 2 to 5 times greater in people with mild to severe HL than in the normal hearing population [5,6].

Over the past two decades, theoretical classes of hypotheses have been developed to define the link between HL and cognitive decline. However, as pointed out by numerous studies, e.g., refs. [7–9], we have still not clearly defined the mechanisms and direction of this link, despite remarkable progress in neuropsychology and neuroaudiology. With the aim of providing a concise theoretical framework, possible causal and non-causal mechanisms for the association between HL and cognitive decline are summarized in Table 1a,b.

Table 1. Causal and non-causal mechanisms linking HL and Cognitive Decline.

(a) Main Hypotheses for the Relationship between HL and Cognitive Decline	
Cognitive Load Hypothesis [4]	The cognitive load hypothesis suggests that HL leads to greater sensory-perceptual effort because of the incoming degraded auditory signal. The greater cognitive resources required for auditory perceptual processing have negative effects on cognitive, attentional, and mnemonic resources. In other words, cognitive decline in hearing-impaired adults might be a consequence of an overinvestment of brain activity in auditory and spoken language processing, resulting in a significant detriment to other cognitive processes.
Information Degradation Hypothesis [10]	The "information degradation hypothesis" suggests that degradation of stimuli (noisy environment, decrease in auditory sensitivity) requires an additional effort: as a consequence, cognitive resources used for signal codification are not available for cognitive functions.
Sensory Deprivation Hypothesis/Cascade Hypothesis [6,11]	According to the "sensory-deprivation hypothesis", HL demands increased cognitive effort which results in depleting cognitive performance over time. Subsequently, cognitive performance deterioration leads to social isolation that in turn causes gradual cognitive decline. Cognitive decline is believed to be potentially remediable with rehabilitation.
Common Cause Hypothesis [12]	Presbycusis and cognitive impairment might be signs of a common neurodegenerative process. So, sensory functioning could be a strong late-life predictor of individual differences in intellectual functioning and could be seen as an indicator of the physiological integrity of the aging brain.
(b) Possible non-causal mechanisms linking HL to cognitive decline	
Testing bias [7–10]	Poor verbal communication associated with HL may confound cognitive testing. HL may influence neuropsychological testing more than cognition per se. HL may introduce a systematic bias into neuropsychological assessments that are mostly designed and validated for verbal instructions and/or the presentation of stimuli. Greater sensitivity of tests in one domain (hearing or cognition) could identify deficits in that domain prior to the other one, leading to the appearance of an illusory causal relationship.
Conceptual bias [7–10]	Upstream common causes with no conditions causally related to others. HL brings older adults to medical attention more often.

For the present study, the "Cognitive Load Hypothesis" (see Table 1a) is of particular interest. According to this hypothesis, the extra effort required for auditory sensory/perceptual processing might be a significant cause of faster cognitive decline in hearing-impaired adults. More specifically, a remarkable auditory processing deterioration for spoken language is one of the issues that occurs most frequently with aging. Moreover, age-related HL entails a perceptual decline in acoustic discrimination of time, intensity, and frequency domains, specifically critical for recognition of words, as well as a dysfunctional central auditory integration, specifically critical for sound localization and recognition of spoken language in a noisy environment. Indeed, difficulties in following a conversation in noisy listening situations represent a typical manifestation of age-related HL [13].

The growing evidence reflecting HL as a controllable risk factor for dementia has supported the research aiming to unveil the potential benefit of aural rehabilitation in cognitive performance. Conventional auditory rehabilitation offers hearing aids for moderate to severe HL and cochlear implants (CI) for severe to profound HL [8,11,14]. Regardless of age, CIs constitute the most suitable and valid auditory prosthetic solution to restore functional hearing in people with severe to profound sensorineural HL [15,16].

The effect of hearing restoration on cognitive functions in CI users is a relatively new topic. Indeed, until 2015, there was a lack of prospective studies on the assessment of postoperative cognitive functions in elderly CI users [17]. However, in the last seven years, several studies have investigated this topic, e.g., refs. [3,14,16,18–23]. These studies found controversial results: some of them indicated postoperative cognitive improvement, e.g., ref. [3], while some others did not observe any significant performance improvements, e.g., ref. [22]. A review by Claes et al. [24] reported that the majority of the existing studies (five out of six) resulted in a significant postoperative cognitive benefit [18–20,25,26] whilst only one study did not observe any significant performance improvement [21].

Conversely, the efficacy of CIs in speech perception has been widely studied and recognized. It is considered the best practice for a wide range of ages, including the elderly population, although the extent of the benefit is highly variable [27]. Among predictive factors, demographic (older age at implantation), audiological (duration of HL, decline in spectral resolution and sensitivity to temporal cues, the amount of preoperative/postoperative residual hearing), and surgical factors (positioning of the electrode array and the angle of insertion) [27–30] seem to account for 10 to 20% of the variability in CI outcomes [31].

The role of neuropsychological functions in elderly CI outcomes has been emphasized by a growing number of studies and their findings might be crucial for a deeper understanding of the link between HL and cognitive decline, e.g., [1,14,23,27,30,32–34]. At present, the outcomes from these studies do not offer a basis for solid conclusions owing to multiple factors. Among them, two factors that are very important for the goals of the present study need to be discussed in detail here.

The first factor is linked to the construct of "cognition" and "cognitive skills" [35] as well as to the variability in the assessment tasks. Cognition is "information processing" and encompasses several functions of different complexity, ranging from subcortical stimuli processing to basic (attentional control and memory) and higher-order executive functions [35]. Indeed, cognition is a very complex construct, an "umbrella term" including several domains and subdomains. Hence, the results of studies on this topic should be interpreted by considering their conceptual and methodological framework.

In the context of research on CI outcomes, the most studied cognitive function has been working memory (WM): there is broad consensus that verbal WM is a cognitive function significantly involved in speech recognition by normal hearing and hearing-impaired subjects [36–39], and that its capacity declines with aging [27,40]. WM is a dynamic memory system that is able to temporarily store and process various information, necessary for complex cognitive tasks such as comprehension, learning, reading text, problem-solving, and reasoning [41]. Despite the presence of various WM conceptualizations with different theoretical and research proposals, most models recognize WM as a dual mechanism consisting of short-term storage and an information processing component [27]. Audiological researchers investigating the role of WM in speech recognition have used a wide variety of tasks for the assessment of the same construct, ranging from less demanding tasks (e.g., a forward digit span) to more demanding ones (e.g., a backward digit span or reading span task). These tasks differ from each other significantly, as they focus on different aspects of cognitive functioning. As underlined by Moberly et al. [27], a forward digit span test primarily assesses the storage component of WM (*how many elements are you able to recall?*) whereas a backward digit span involves more processing components (*how many elements are you able to recall in inverse order?*) [42]. Hence, the results of WM studies in hearing-impaired people may partly depend on the theoretical framework and the type of task. A second factor is the difficulty of isolating one cognitive domain from another when studying their effects on individual CI outcomes. Among cognitive domains, WM and its role in speech recognition outcomes has been the most common research topic [27]. Especially in noisy listening contexts, WM compensates for the degraded auditory signals provided by amplification systems and/or CI [34]. However, it should also be considered that WM tasks always involve some level of attention [40,43] and so far, the impact of attention on WM performance might have been ignored. Attention is not a unitary concept [44] but a complex construct of various definitions and conceptualizations [43,44]. Nevertheless, in general terms, attention can be defined as the ability of our perceptual system to select the information of interest to us within our processing capacity, acting as an input filter [45]. In this sense, attention is seen as a mechanism of "priority selection" for the information to be processed, e.g., ref. [44]. From another theoretical perspective, attention is defined as a limited resource of information processing [43]. Theories conceptualizing attention as a resource assume that this resource is responsible for the limited capacity of WM [43] and this association between WM and attention is also referred to as executive attention [46].

General cognition, WM, and attention are conventionally considered predictors of better CI performance [18,27,34]. In clinical and experimental contexts, the difficulty of disentangling the assessment of one cognitive domain from another presents itself at any age but becomes even more evident in elderly people with sensorineural HL. This is mainly due to the impairment of contextual hearing and the aging of the brain, associated with slower cognitive processing and a decline in attentional resources [47]. Thus, the present study aimed to investigate some specific cognitive domains in elderly CI users and to try to disentangle the effects of these cognitive skills on speech perception. Considering HL as a significant risk factor for cognitive decline [5,6,11], a better understanding of audiological aspects in the elderly CI population could enable early/special intervention of cognitive functions, providing a potential benefit for postoperative cognitive and audiological outcomes [3,48].

2. Materials and Methods

2.1. Participants and Study Design

The present study consisted of elderly CI users, all implanted and regularly followed at the CI center of the Sapienza University of Rome (Policlinico Umberto I). Informed consent was obtained from each subject prior to study enrollment, and all procedures were approved by the local ethics committee of the Sapienza University of Rome (Protocol no: 5982, 22.04.2020).

The inclusion criteria for study enrollment were being ≥60 years of age and having >12 months of CI experience. The data collected included age at implantation, gender, side of implantation, listening mode (unilateral, bilateral, or bimodal), duration of HL, etiology, aided sound-field (SF) audiometry, speech perception assessment in both quiet/noise, and cognitive performance in a memory, attention, and reasoning test. The exclusion criteria regarded as significant a self-reported history of psychiatric conditions and/or diagnosed incident dementia as well as a cognitive and anxiety level outside the normal clinical range. Elderly CI users with any comorbid medical conditions potentially impacting neuropsychological functioning including stroke, ischemic attack, traumatic brain injury, and concussion were excluded from this study.

Thirty postlingually deafened CI users, all native Italian speakers, participated in the present study. All unilaterally and bilaterally implanted (UCI and BCI, respectively) participants had bilateral severe to profound HL. Bimodal users (CI and a contralateral hearing aid) (BIM) showed severe to profound HL in the implanted ear and a down-sloping moderate to severe HL on the hearing aid side.

All 30 participants successfully completed the cognitive assessment battery; hence, their data were all included in the statistical analysis. Participants (16 M and 14 F) had a mean age of 73.4 (range 60 to 87 years; SD = 6.6). Twenty participants (66.7%) had a basic education (primary and lower secondary); eight (26.7%) had an intermediate upper secondary level, and two (6.7%) had an advanced educational level (bachelor's or equivalent degree).

The mean duration of HL—defined by self-reported first hearing aid use—was 36.7 years (range 6 to 70 years; SD = 16.4) and the mean duration of CI experience was 8.6 years (range 1 to 22 years; SD = 5.54). A total of 15 participants (50%) were unilateral CI users, whilst 5 participants (16.7%) had bilateral CI and 10 participants (33.3%) were bimodal (CI/HA) users. A total of 12 participants had AB® cochlear implants (1 CII HF 1J, 5 HiRes 90K adv, 3 HiRes 90K HF1J, and 3 HiRes Ultra MS receivers; all using Naida 90 BTE Processors) fitted with Optima™ strategy; 14 participants used Med-El devices (1 Pulsar CI-100 and 13 Synchrony receivers; all using Sonnet BTE Processors) fitted with FS4 strategy; 4 received Cochlear® devices (2 CI24RE, 1 CI512, and 1 CI632 receivers; all using Nucleus 7 BTE Processor) fitted with ACE™ strategy. The descriptive data of the participants are shown in Table 2.

Table 2. Descriptive data of the participants (n = 30).

Personal Variables		Mean (sd)
Age at test (years)		73.4 (6.6)
Duration of HL (years)		36.7 (16.4)
CI experience (years)		8.6 (5.54)
		n (%)
CI listening mode	Unilateral	15 (50.0)
	Bilateral	5 (16.7)
	Bimodal (CI/HA)	10 (33.3)
Gender	Male	16 (53.3)
	Female	14 (46.7)
Status	Married	23 (76.7)
	Unmarried	2 (6.7)
	Widow	5 (16.7)
	Living alone	4 (20.0)
	Living with significant others	16 (80.0)
Educational level	Basic	20 (66.7)
	Intermediate	8 (26.7)
	Advanced	2(6.7)

Abbreviations: CI = cochlear implant; HA = hearing aid.

2.2. Procedure

The procedure consisted of an audiological evaluation followed by a cognitive assessment.

2.2.1. Audiological Assessment

An audiological assessment was performed with participants' everyday listening mode. Pure tone SF audiometry was measured both for unilateral and bilateral/bimodal listening conditions. The assessment was performed in a sound-proof audiometric chamber using an Aurical audiometer (Otometrics Taastrup, Denmark) connected to a loudspeaker placed at 0° azimuth at 1 m distance from the participant's head.

Speech perception tests consisted of Italian disyllabic words (W) and sentences (S) [49]. Tests were administered in quiet (Wq/Sq) and in noise with a fixed signal-to-noise ratio (SNR) at +10 (W+10/S+10) and +5 dB (W+5/S+5). Both speech and noise signals were presented from a loudspeaker at 0° azimuth at a 1 m distance from the participant's head and the primary signal was fixed at 65 dB HL. Testing for each speech material was preceded by a training list.

2.2.2. Cognitive Assessment

All participants were tested individually in a quiet room by a psychologist experienced in the clinical assessment of hearing-impaired patients. To facilitate speech comprehension during the assessment procedure, the psychologist did not wear an FFP-2 mask, as indicated by government provision. The cognitive assessment was performed in everyday best-aided listening mode.

For the screening of general cognitive functioning and anxiety symptoms, the following assessment tools were used: the Raven Colored Progressive Matrices (CPMs) [50,51], the Montreal Cognitive Assessment [52,53], and the State-Trait Anxiety Inventory for Adults (STAI-Y) [54]. The CPMs have been designed to evaluate the cognitive level of children (with typical/atypical development) and adults with intellectual disabilities or elderly with cognitive decline. The CPMs' normal values range between 25° and 75° percentile and the risk of dementia is considered at <5° percentile. The verbal (culturally based) cognitive functioning was assessed via the Montreal Cognitive Assessment (MoCA) [52]. Normative values are based on an Italian adult population [53]: the test considered a cut-off value of 15.5 (corrected for age and educational level) to be indicative of a significant cognitive decline. The State-Trait Anxiety Inventory for Adults (STAI-Y) [54] is a well-known and easy-to-apply questionnaire for the detection of anxiety symptoms in adult populations. The range of possible scores for each scale varies from 20 to 80, with a predictive threshold value of severe anxious symptomatology set at 60.

Aside from the screening, a psychometric test battery was implemented to assess short-term memory, verbal WM, attention domains, and state-trait anxiety symptoms. The cognitive assessment battery consisted of the following:

Forward and backward digit span (FDS and BDS): these are the most frequently used tests of short-term memory/simple WM [55]. In the present study, we used the version of De Beni and Borella [56]. The test consists of two tasks: a forward digit span (passive short-term memory) and an inverse digit span task (active short-term memory). The examiner verbally presents digits at a rate of one per second. In the *forward form*, the participant is required to repeat the digits verbatim. In the *backward form*, the participant has to repeat the digits in reverse order. The score is corrected for age, gender, and educational level.

Categorization working memory span task (CWMT) [56]. This task was used to assess participants' verbal WM. It consists of 20 lists of 5 words (100 words in total) organized in sets of 3 to 6 lists. Each list contains a maximum of 2 animal names. The participant is required to listen to the lists, memorize the final word of each list, and remember the words in the last position with the correct order of presentation at the end of each set. In addition to the memory task, the participant is required to perform a secondary task which is to beat the hand on the table every time the name of an animal appears. The total number of correctly remembered words represents the verbal WM capacity index (maximum score = 20) [56]. The score is corrected for age, gender, and educational level.

RBANS attention subsection: The evaluation of attention capacities was carried out through the administration of subtests from the repeatable battery of assessment of neuropsychological status (RBANS) [57]. The RBANS attention domain consists of two subtests (1 verbal digit span and 1 nonverbal symbol/digit association): the combination of the obtained raw score provides an attention domain index score which is corrected for age at the time of the test. Following the recommendations of Patton et al. [58], who applied cut-offs to the RBANS score to increase scale sensitivity, attention performance was analyzed as standard scores and subsequently, performances were divided into two subgroups: low/medium attention performers (LMAP) ($\leq$90) and high attention performers (HAP) ($\geq$91).

2.3. Statistical Analysis

Descriptive and inferential statistical analyses were performed. After checking the normality of each data distribution with both Shapiro–Wilk and Kolmogorov–Smirnov tests, a non-parametric analysis was adopted. Kruskall–Wallis and Mann–Whitney U tests (with Bonferroni correction for multiple comparisons, when necessary) were used to compare the effects of listening mode (UCI, BCI, and BIM) and gender on cognitive (FDS, BDS, CWTM, RAS, STAI-Y-1, and STAI-Y-2) and audiological (SF and speech perception) outcomes. The statistical analysis showed no statistically significant effects ($p > 0.05$).

Therefore, the participants were not divided into subgroups with respect to the variables of gender and listening mode. Subsequently, Mann–Whitney U tests were performed to compare participants based on attention performance (LMAP and HAP based on a cut-off of 90) [56]. The effect size was calculated using the Rosenthal formula r1/4Z/N (small effect 1/4 0.10–0.30, moderate effect 1/4 0.30–0.50, and large effect >0.50) [59].

For statistical analysis, the percent correct scores for speech perception in quiet were transformed into rationalized arcsine units (RAUs) to avoid the ceiling effects [60].

Finally, a Spearman correlation analysis was performed to evaluate the associations between cognitive variables, and a simple regression analysis investigated the relationships between cognitive and audiological variables; p-values ≤ 0.05 were considered statistically significant. For multiple comparisons (SF thresholds and word/sentence recognition) with Bonferroni correction, $p \leq 0.017$.

3. Results

3.1. Outcomes of Attention Assessment and Their Comparison with Audiological/Cognitive Data

The median attention values for the LMAP and HAP subgroups were 74 (range 58 to 89) and 100 (range 90 to 126), respectively. The median SF threshold for octave frequencies between 250 to 4000 Hz was 31 dB HL (range 20 to 55 dB HL). The corresponding value was 35 dB HL (range 20 to 50 dB HL) at 250 Hz, 30 dB HL at 500 Hz (range 20 to 40 dB HL), 30 dB HL at 1000 Hz (range 20 to 45 dB HL), 30 dB HL at 2000 Hz (range 20 to 45 dB HL), and 35 dB HL at 4000 Hz (range 20 to 55 dB HL).

A total of 93% of subjects showed a score > 50% for word and sentence recognition in quiet. Word recognition scores from the LMAP and HAP subgroups were: 80% (22 to 100%) versus 90% (range 58 to 100%) in quiet, 40% (range 0 to 55%) versus 60% (range 0 to 90%) at SNR+10, and 0% (range 0 to 20%) versus 28% (range 0 to 88%) at SNR+5. Sentence recognition scores from the LMAP and HAP subgroups were: 90% (range 0 to 100%) versus 90% (range 70 to 100%) in quiet, 40% (range 0 to 70%) versus 60% (range 0 to 100%) at SNR+10, and 0% (range 0 to 10%) versus 20% (0 to 100%) at SNR+5.

For the cognitive variables, the comparison between the LMAP and HAP subgroups showed statistically significant differences for both CWTM and DIGIT performance. Participants with high attentional performance had significantly higher values than low performers for all WM tests (Table 3). Except for CWTM (effect size = 0.42), all statistically significant differences showed a medium/large effect size (0.5–0.64).

3.2. Correlations and Regressions

Spearman's correlation analysis was carried out to see which sociodemographic and audiological data were significantly related to cognitive variables (Table 4). Strong correlations were observed between attention and all other cognitive variables ($0.534 \leq \text{Rho} \leq 0.742$, $p < 0.01$). Statistically significant correlations were found between attention and both SF ($-0.404 \leq \text{Rho} \leq 0.581$, $p < 0.01$) and speech perception in noise outcomes ($0.397 \leq \text{Rho} \leq 0.664$). Interestingly, the results showed a statistically significant correlation between attention and educational level (Rho = 0.604) whereas for CWTM significant correlations were observed only with W+10 and W+5 (Rho of 0.497 and 0.519, $p < 0.05$, respectively).

Table 3. The table shows the differences between the LMAP and HAP subgroups for dependent cognitive variables, measured with the Mann–Whitney U test and adjusted with Bonferroni correction for multiple comparisons. Significant correlations are shown in bold. For multiple comparisons (word and sentence recognition) with Bonferroni correction, p-values were $\leq$0.017 (*).

A. *Cognitive Variables*	**LMAP Rank Sum**	**HAP Rank Sum**	**U**	**Z**	***p*-Value**	**Effect Size**
CWTM	146.000	319.000	55.000	2.322	0.019	0.425
FDS	119.000	346.000	28.000	3.452	0.000	0.642
BDS	137.000	328.000	46.000	2.699	0.005	0.511
B. *Audiological Variables*	**LMAP Rank Sum**	**HAP Rank Sum**	**U**	**Z**	***p*-Value**	**Effect Size**
W quiet	113.50	211.50	47.50	1.632	0.106 *	0.320
W+10	92.00	233.00	26.00	2.391	0.005 *	0.470
W+5	102.50	222.50	36.50	2.798	0.026 *	0.550
S quiet	127.00	198.00	61.00	0.919	0.381 *	0.180
S +10	112.50	212.50	46.50	1.685	0.094 *	0.330
S +5	126.00	225.00	48.00	2.017	0.064 *	0.400

Abbreviations: LMAP = low–medium attention performers group (RBANS attention subscale < 90); HAP = high attention performers group (RBANS attention scale $\geq$90); RBANS = repeatable battery of assessment of neuropsychological status; CWTM = categorization working memory span task; FDS = forward digit span; BDS = backward digit span; W = words, S = sentences; +5, +10 = signal-to-noise ratio.

For the whole group, the simple linear regression analysis with Bonferroni correction showed a statistically significant linear dependence between attention and aided SF at 2000 Hz ($R = 0.611$, $R^2 = 0.379$, $p = 0.005$) and 4000 Hz ($R = 0.753$, $R^2 = 0.567$, $p \leq 0.0001$) as well as a trend toward significant linear dependence with aided SF at 250 Hz ($R = 0.489$, $R^2 = 0.235$, $p = 0.035$). Attention significantly predicted speech perception in noise for W+10 (SNR+10: $R = 0.530$, $R^2 = 0.281$, $p = 0.017$) (Figure 1) while there was a trend of significance for W+5 (SNR+5: $R = 0.538$, $R^2 = 0.289$, $p = 0.019$) and sentences (SNR+5: $R = 0.470$, $R^2 = 0.221$, $p = 0.04$; SNR+10: $R = 0.470$, $R^2 = 0.221$, $p = 0.04$) (Figure 2). Simple linear regression based on the RBANS score explained more than 50% of the variance for word recognition in noise and more than 46% of the variance for sentence recognition in noise.

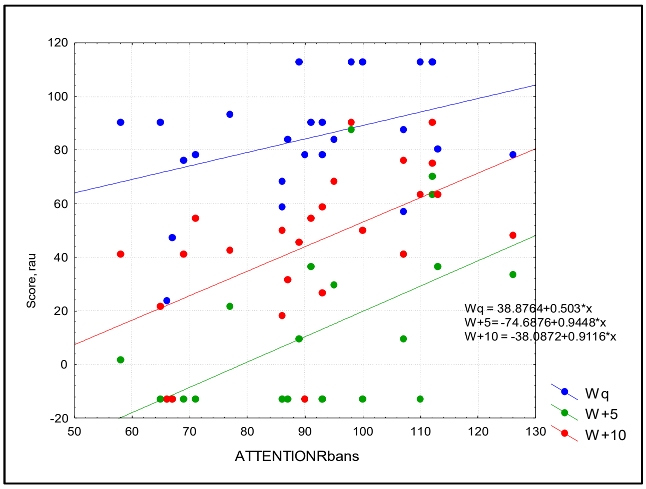

Figure 1. Scatterplot of audiological outcomes for words in three conditions: blue = quiet (Wq), green = SNR = +5 (W+5), and red = SNR+10 (W+10) as predicted by attention in the study sample (n = 30). Simple linear regression based on the RBANS score explained more than 50% of the variance for word recognition in noise.

Table 4. Relationships between sociodemographic, cognitive, and audiological variables in the overall study group (n = 30).

VARIABLES	SF 250 Hz	SF 500 Hz	SF 1000 Hz	SF 2000 Hz	SF 4000 Hz	Wq	W+10	W+5	Sq	S+10	S+5	Age at Test (yrs)	Educa-tion (yrs)	HL duration (yrs)	Atten-tion	CWTM	FDS	BDS
Age at test (yrs)	**0.43** *	0.32	0.23	0.29	0.15	−−0.25	**−0.47** *	**−0.52** **	**−0.44** *	−0.15	−0.27	−−	**−0.38** *	−0.27	−0.34	−0.30	−0.07	0.002
Education (yrs)	**−0.56** **	**−0.40** *	**−048** *	**−0.42** *	**−0.53** **	0.25	0.38	**0.43** *	0.37	0.1	0.25	−0.37	−−	−0.09	**0.60** **	0.30	0.27	**0.53** **
HL duration (yrs)	0.00	−0.34	−0.15	−0.19	−0.16	0.08	0.32	0.15	0.30	0.45	0.23	−0.27	−0.10	−−	−0.12	−0.00	−0.27	−0.05
Attention	**−0.58** **	**−0.40** *	−0.37	**−0.45** *	**−0.47** *	0.356	**0.66** **	**0.58** **	0.32	0.34	**0.40** *	−0.34	**0.60** **	−0.19	−−	**0.67** **	**0.74** **	**0.53** **
CWTM	**−0.50** *	**−0.41** *	−0.33	−0.38	−0.28	0.31	**0.50** *	**0.52** *	0.07	0.28	0.33	−0.31	0.31	−0.00	**0.66** **	−−	**0.56** **	0.32
FDS	**−0.40**	−0.29	−0.32	−0.32	−0.27	0.06	**0.40**	0.38	−0.12	0.25	0.27	−0.07	0.27	−0.27	0.74	**0.56** **	−−	**0.46** *
BDS	−0.31	−0.36	**−0.41** *	−0.39	**−0.54** **	0.07	0.27	0.25	−0.06	0.27	0.22	0.00	0.53	−0.05	**0.53** **	0.32	**0.46** *	−−

* = correlation (Rho) statistically significant at $p < 0.05$; ** = correlation (Rho) statistically significant at $p < 0.01$. Significant correlations are in bold. Abbreviations: HL = hearing loss; CWTM = categorization working memory span task; FDS = forward digit span; BDS = backward digit span; SF = sound-field audiometry; W: words, S: sentences, q = quiet condition; +5 = signal-to-noise ratio +5; +10 = signal-to-noise ratio +10.

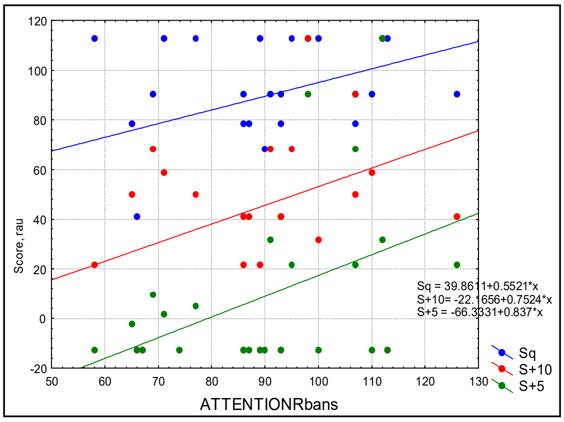

Figure 2. Scatterplot of audiological outcomes for sentences in three conditions: blue = quiet (Sq), red = SNR = +10 (S+10), and green = SNR+5 (S+5) as predicted by attention in the study sample (n = 30). Simple linear regression based on the RBANS score explained more than 46% of the variance for sentence recognition in noise.

Following post hoc analysis, WM showed a trend toward a significant prediction of W+10 (R = 0.43, $R^2 = 0.19$; $p = 0.031$) and W+5 (R = 0.47, $R^2 = 0.224$; $p = 0.019$) (Figure 3).

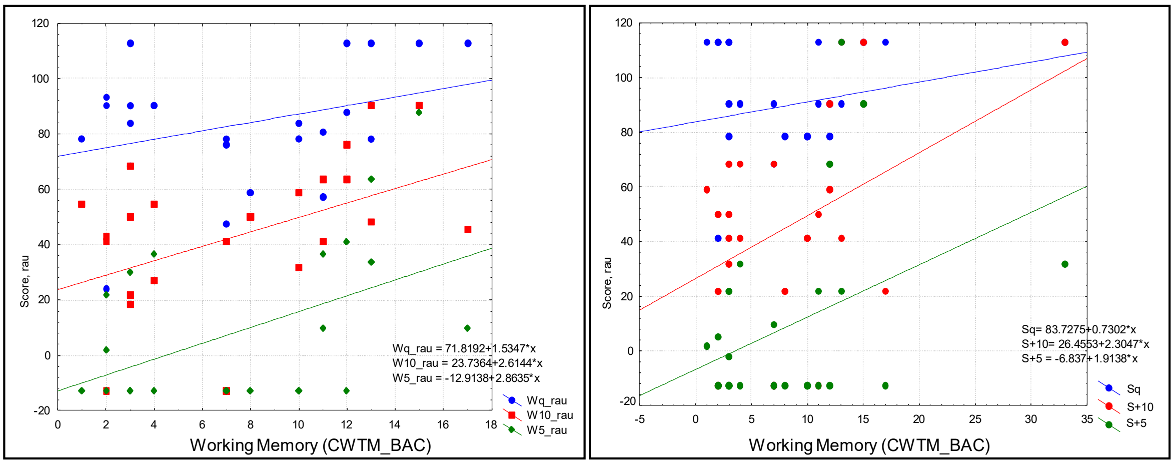

Figure 3. Scatterplot of audiological outcomes. Left: words in three conditions (blue = quiet (Wq), red = SNR = +10 (W+10), and green = SNR+5 (W+5)). Right: sentences in three conditions (blue = quiet (Sq), green = SNR = +10 (S+10), red = SNR+5 (S+5)) as predicted by WM in the study sample; scatterplots were produced without the contribution of outliers in the analysis: W+10 (R = 0.43, $R^2 = 0.19$; $p = 0.031$) and W+5 (R = 0.47, $R^2 = 0.224$; $p = 0.019$).

4. Discussion

Cognitive domains such as attention, cognition, and WM are believed to contribute significantly to better CI outcomes, especially in postlingually deafened elderly populations, e.g., refs. [14,31–34]. Hence, the present study aimed to gain insight into the effects of these cognitive functions on postoperative speech perception outcomes for an elderly CI population. Not surprisingly, the present findings reflected the significant link between attention, cognition, and WM. Moreover, attention resulted in playing a significant role in outcomes from both tonal and speech audiometry in noise whilst WM was significantly related to speech perception in noise for this sample of elderly CI users. More specifically, the important role of cognitive functions became more apparent for open-set recognition of words in noise than that of everyday sentences, where the increase in speech material's semantic predictability might have provided a remarkable perceptual benefit for reducing the cognitive load and listening effort as attentional effort, despite the challenges of listening in noise [61].

Overall, the findings highlighted that better cognitive performance may positively contribute to better speech perception outcomes, particularly for complex listening situations where noise is present, and performance relies more on the perception of auditory cues such as open-set words instead of highly predictable everyday sentences. Such results were further supported by the present significant correlations between attention and aided SF audiometry, which was observed for the first time specifically in elderly CI users. In other words, higher attention scores predicted lower (better) CI thresholds or vice versa. Similar to a previous paper by Mancini et al. [62], the present sample showed aided SF thresholds in line with those obtained from typical adult CI users. Likewise, even for a group of CI listeners who achieved target PTAs better than 40 dB HL, the significant effects of aided SF thresholds on a speech recognition test (the STARR test) presenting everyday sentences at low, medium, and high levels in adaptive noise were reported by Dincer D'Alessandro et al. [63]. Moreover, better STARR findings were observed for a group of elderly bimodal users, benefiting from the summation effect to improve overall bilateral audibility when listening both with a CI and a contralateral hearing aid [62]. Indeed, perceptual difficulties for speech at the low level, probably increasing listening effort linked to degraded auditory input requiring greater auditory attention as in the case of shorter duration and less predictable word recognition test in noise here, appeared to be the major factor limiting STARR performance despite the highly predictable everyday sentences of the test [63,64]. Similarly, a significant difference between low and high attention performers for word recognition at SNR +10 was found in the present study group. Despite the increased number of CI users showing floor effects for word recognition at SNR + 5, a trend toward a significant difference was observed for this test condition as well. Hence, it is reasonable to expect a significant difference for a larger population instead of the present LMAP (n = 13) versus HAP (n = 17) subgroup comparisons with a smaller sample size. Indeed, the use of Bonferroni correction in such a small sample might have been too conservative due to the risk of Type 2 statistical error linked to an increased probability of producing false negatives [65]. Such an expectation is also supported by a good correlation index found by bivariate correlational analysis, reflecting statistically significant correlations between attention and speech perception in noise for both words and sentences in the overall group (n = 30). Despite losing significance after the Bonferroni correction, a trend toward significant dependence was still present for the linear regression analysis as well.

Recently, the effects of cognition and attention on speech perception performance have been emerging topics in the field of cochlear implantation. However, the existing literature is limited to a few studies. This fact might partly stem from the limited number of available tools to assess such specific cognitive functions specifically in elderly hearing-impaired populations. There are a few screening tools, such as the RBANS and MoCA, used for the assessment of cognitive domains, including cognition and attention in relation to auditory skills in elderly CI recipients. The RBANS is a well-known neuropsychological tool for clinical diagnosis and follow-up of dementia and mild cognitive impairments. The test offers a total score of cognition, which can also be divided into five cognitive domains, including attention. A newer version of this tool, RBANS-H, has been specifically developed for hearing-impaired people, with the aim of avoiding the bias linked to HL, especially in the elderly population [66]. On the other hand, the MoCA is a rapid screening tool to measure cognition and addresses questions regarding memory recall and executive functions. The tool evaluates components concerning delayed memory, visuospatial abilities, executive functions, language, attention, and orientation [52].

Unlike the present work, previous studies using RBANS-H [3,67] and MoCA [68] did not observe any significant correlations between attention and speech perception in noise. Such outcome differences might be partly owing to differences in study samples and methodology. More specifically, the present work used the original version of RBANS instead of RBANS-H designed specifically for hearing-impaired people. The RBANS attention task adopted in the present study consists of a nonverbal symbol/digit association and a verbal digit span—this last one was administered in auditory–visual mode with available lipreading cues. Concerning the attention task, both the RBANS-H and RBANS have the same nonverbal symbol/digit association, while the digit span is administered in an auditory–visual modality in the case of RBANS-H. On the other hand, Vasil et al. [68] study used MoCA subscales to investigate correlations with speech perception outcomes. The authors found a significant correlation between the percentage of correct recognition scores for words and the test of delayed recall, for which stimuli were presented in an auditory-alone condition. However, they did not observe any significant correlations with the attention subtests which are administered in auditory-alone conditions as well. The authors speculated that correlations and linear regression models might be interpreted in two ways: cochlear implantation may improve the performance of cognitive functions even for nonauditory stimuli; or, alternatively, individuals with better cognitive skills show better postoperative speech perception performance. In light of the present findings, we believe that differences in correlations between the two auditory tasks might be partly dependent on the cognitive domain, and significant correlations between attention and auditory perception may support the hypothesis that individuals with better attention skills might be those who make better use of auditory cues conveyed by CI and consequently show better postoperative performance.

The present sample was divided into two subgroups of low and high performers (LMAP and HAP) with the aim of gaining insight into the role of attention in other cognitive functions. These subgroups showed statistically significant differences for both the categorization of WM and forward/backward digit span tests. More specifically, participants with high attentional performance had significantly higher values than low performers for all cognitive tests. Moreover, attention performance was highly correlated with WM, indicating a medium/high coefficient. This result appears highly consistent with the existing literature [43,44,46]. There is a strong link between WM and attention [43], and thus, it is possible to state that "by virtue of holding a selected subset of all available representations in memory, WM is by definition a form of attention" [43], p. 14. A main strand of empirically supported theories has conceptualized attention as a limited resource for information storage and processing, suggesting attention to be responsible for the limited capacity of WM [43]. Interestingly, a different strand of theories also defined attention as a selection process [69]. In this sense, WM can also be conceptualized as an "instance of attention" [43], p. 7: whilst selective attention is "attention to perceptual objects", WM is "attention to memory objects". Finally, WM capacity is believed to be closely related to attention ability in order to focus on a target by excluding any distractor from the encoding process [40]. Indeed, several researchers support the hypothesis that WM capacity might be limited by an attentional resource [43]. Unlike younger adults, in the elderly population, a higher reliance on attention skills might be required for encoding a WM task without distraction.

Age-related cognitive decline negatively affects speech recognition, especially in challenging situations such as listening in the presence of competing noise [6,11]. Therefore, in a complex auditory scenario, cognitive resources are spent on perceptual processing to the detriment of other cognitive processes [70,71]. Hence, it is conceivable that in the HAP subgroup—because of higher attentional resources—the cognitive load needed for storing and processing information might be lower than in the LMAP subgroup, significantly influencing the performance for both simple and complex WM tasks.

As mentioned above, according to the present findings, attention seemed to play a significant role in speech perception, especially for more complex listening tasks: the HAP subgroup showed significantly better outcomes in the perception of words presented in noise (W+10). Similarly, regression analysis showed that attention significantly predicted recognition of words presented at SNR+10. Moreover, for the linear regression analysis of words presented at SNR+5 and sentence recognition in noise, the presence of a trend toward significance even after Bonferroni correction in such a small sample was a promising finding. Indeed, it is a well-known fact that "top-down" cognitive processes are key elements of speech perception, linking incoming acoustic signals to phonological and lexical representations in long-term memory [27,72]. Nevertheless, cognitive mechanisms involved in speech processing follow different paths depending on whether the incoming sounds are clear (as it happens for normal hearing people and/or in quiet listening situations) or degraded (as it happens for people with sensorineural HL even in the best-aided listening condition, especially in noisy contexts). In fact, current CI technology provides useful acoustic cues in the time, intensity, and frequency domains and a good representation of envelope cues that are required for speech understanding in quiet, whereas temporal fine structure cues, known to be crucial for complex listening such as speech in noise and music, are mostly removed from CI speech processing [13]. As a matter of fact, speech processing strategy is an extremely important aspect of CI technology. Currently, there are various signal processing techniques that differ between CI systems (for more information on this topic, see for example Choi and Lee [73]). Nevertheless, electrical stimulation induces a pattern of auditory nerve activity poorer than acoustic stimulation [74], in particular for coding of low-frequency signals such as the fundamental frequency of voice [75].

According to the ease of language understanding model (ELU) [39], when acoustic signals are clear, incoming sounds are encoded by an implicit effortless speech processing channel. On the contrary, when incoming signals are degraded, perceptual ambiguity increases, and recognition of acoustic information gets harder. In that case, the incoming sounds are encoded by a compensatory, slower, and more effortful explicit channel, that strongly relies on WM. Here, high attention resources are needed to overcome the mismatch between the incoming signal and the encoded one [31,76]. Hence, it is conceivable that better CI performers in terms of speech perception make more robust use of the explicit channel "which requires good working memory and a high level of attention to match the degraded signal with the storage in the long-term memory" [31], p. 549.

Whilst the link between WM and CI outcomes has been widely studied [16,31,34,42,77], the role of attention in CI outcomes has received less attention. Very recently, auditory selective attention has been shown to significantly affect linguistic outcomes in pediatric CI users [78]. Auditory attention largely depends on listeners' ability to enhance the representation of a target auditory source. This requires analyzing the acoustic scene and segregating the target sounds, showing attentional focus on the target, suppressing interfering elements, and simultaneously maintaining cognitive flexibility to switch attention toward new auditory targets required by the context [79]. It is a complex process at any age in the hearing population but becomes even more effortful in CI users, because of the distorted transmission of auditory objects, especially in a noisy environment [78]. It is conceivable that in our sample of elderly CI recipients, higher attentional resources may lead to diminishing cognitive resources required to store and process auditory information (WM), which is especially important for understanding complex speech. In this light, higher attentional resources may allow using attentive skills both for auditory processing and memory representations. Conversely, low attentional resources in the LMAP subgroup might be a significant cause of lower scores for word recognition in noise (both at SNR +5 and +10 dB), where an increased cognitive load is needed to fill the perceptive gaps left by inaudible parts in the acoustic streams [80]. This observation is in line with a study by Volter et al. [31] showing that differences in cognitive functioning and linguistic skills may explain poor speech recognition scores in adult CI performers. Although significant differences between poor and good CI performers in terms of speech perception could be detected in various cognitive subdomains, Volter et al. [31] found that the most prominent difference was observed for the attentional task. Attention had the strongest power to discriminate between poor and good performers in elderly CI users, and it was significantly improved after one year of CI use. Similar to Volter et al. [31], in the present study the correlation analysis seems to reflect a significant role of attention in CI outcomes both for tonal and speech audiometry in noise whereas, for verbal WM (CWTM), the only significant correlation was observed with words at SNR+10 and +5. Furthermore, the present findings from regression analysis showing the presence of a trend toward significance even after

Bonferroni correction supported such arguments. These findings, taken together, seem to support the ELU model [39]. Reducing the extent of mismatch between incoming speech stimuli and their phonological representation in long-term memory, CI provides a reduction in attentional effort. In turn, increased availability of attention might be crucial for WM involvement in speech understanding, especially in noisy environments [23]. On the other hand, it is conceivable that CI users with higher attentional resources may achieve better speech understanding as a result of the significant link between attention and WM [23,43].

Finally, a significant impact of schooling was observed for attention, while no significant associations were found between educational level and WM tasks. These results are inconsistent with Volter et al. [23], who found a significant impact of educational level on a WM task, but no significant impact on attention tasks. Nevertheless, the differences in tasks and CI experience between the study groups may explain such differences. As a matter of fact, in the present study, we used a WM task (CWMT) with a score corrected for age and educational level, whereas the attention task score from RBANS is corrected only for age. Indeed, Claes et al. [67], in their cross-sectional study using the RBANS-H, found a significant positive association between the RBANS-H score and educational level in a sample of experienced adult CI users. It is widely recognized that educational attainment is a key component of successful cognitive aging [81,82]. From a socio-economic point of view, a higher level of education leads to increasing involvement in cognitive-demanding occupations and participation in social/leisure activities that are cognitively more engaging and stimulating.

Interestingly, educational level is strictly linked to the construct of the cognitive reserve [83], a latent construct that has recently been added to the list of modifiable risk factors for cognitive decline/dementia and is made up of several factors with three dimensions (education, work, and leisure activities) [2]. The cognitive reserve is a concept based on brain plasticity and refers to the ability of the brain to cope with damage and aging by using pre-existing cognitive processes or compensatory brain networks [84]. Although the cognitive reserve is mostly indexed by education, it is a concept highly based on brain plasticity; hence, it is made of potentially controllable indicators such as occupational attainment and stimulating activities, e.g., reading, writing, playing music, social engagement, and cognitive exercises [16]. In this light, the construct of the cognitive reserve and the association that we found between attention and educational level might be promising indicators for cognitive interventions in elderly CI users who are potentially able to exploit their residual brain plasticity [85].

5. Limitations

A limitation of the present study stems from its methodology, making it impossible to state a causal relationship among the study variables. Moreover, the absence of preoperative cognitive evaluation does not allow us to investigate the cognitive evolution of the present long-term CI users and the effects of the amount of auditory benefit on their cognitive performance.

Another limitation might be a bias linked to the use of RBANS for assessing attention skills in this group of elderly CI users. RBANS-H might have been a better alternative as it is specifically designed for hearing-impaired people to minimize the effects of bottom-up auditory processing on attentional outcomes. However, it is a matter of fact that there is no adaptation of the RBANS-H in the Italian language. On the other hand, auditory verbal tasks may have a greater relevance to the recognition of degraded speech [27,42,78,86]. Indeed, the present sample consisted of experienced adult CI users, all postlingually deafened. Most of them had a word/sentence recognition score in quiet equal to or greater than 80%, and they were allowed to make use of lipreading cues during testing. None of the participants had a history of auditory deprivation in childhood, and their knowledge of the native language appeared to be comparable to that of a hearing adult of the same age. Hence, auditory verbal instructions presented in a silent environment by a clinician experienced in working with CI users might not be a significant bias. However, without comparisons of RBANS and RBANS-H outcomes in such a population, we cannot rule out a bias linked to bottom-up processing, potentially leading to an underestimation of cognitive functioning [31].

6. Conclusions

The present study observed significant associations between attention and auditory outcomes for both aided SF thresholds and speech perception in noise. Moreover, participants with high attentional performance showed significantly higher scores than low attentional performers for all WM tests. The enhancement of WM and attention skills may positively contribute to speech perception performance in elderly CI users. Whilst WM plays a significant role in the storage and processing of auditory verbal stimuli, more robust attention may lead to better performance for speech perception in noise where a high level of auditory selective attention is needed. In view of the present results, the effectiveness of an early intervention based on auditory cognitive training should be investigated in order to improve cognitive and audiological performance in elderly CI users.

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RESEARCH ARTICLE

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School wellbeing and psychological characteristics of online learning in families of children with and without hearing loss during the Covid-19 pandemic

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Abstract

This study investigated the psychological characteristics of online learning on Italian students with and without hearing loss (HL) and on their parents, who were forced into isolation during the Covid-19 pandemic. An online survey collected information on socio-demographic data and opinions concerning online learning from 61 children (mean age 11; 25 males, 36 females), including 43 with HL and also from their parents; additionally, school wellbeing and anxiety were assessed. The results showed that, in both the student and parent groups, no significant effect of HL on school wellbeing and anxiety was found. Additionally, in parents, State Anxiety was significantly higher than Trait Anxiety, suggesting one possible impact of lockdown on psychological wellbeing. Differences due to HL were observed and discussed in correlation analyses. The Authors believe that this study is the first contribution to the psychological evaluation of the impact of online learning on families with hearing-impaired children, from the perspective of a successful, inclusive didactic.

Giulia Cartocci and Patrizia Mancini equally supervised the project.

KEYWORDS
hearing loss, online learning, school wellbeing

1 | INTRODUCTION

On 12 March 2020, World Health Organization (WHO) declared the coronavirus disease 2019 (Covid-19) outbreak to be a pandemic (World Health Organization, 2020). Numerous countries have instituted large-scale or national school closures to decelerate virus transmission by encouraging social distancing (Viner et al., 2020). As a result, in Spring 2020, many educational institutions decided to move from in-person instruction to a remote learning model (Di Pietro et al., 2020). Online learning can be defined as "learning experienced through the internet/online computers in a synchronous classroom where students interact with instructors and other students and are not dependent on their physical location for participating in this online learning experience" (Singh & Thurman, 2019; p.302). Despite a plethora of technologies available for online education, complex student challenges can occur (Dhawan, 2020; Song et al., 2004). Researchers (R. T.-H. Chen, 2010; O'Doherty et al., 2018) expressed their concerns about online learning and highlighted the main difficulties associated with creating an online learning community involving higher levels of social presence and engagement. In addition, scholars have concerns about social isolation, the lack of interactivity and participation and delayed or insubstantial amounts of feedback (Dong & Mertala, 2021; Khurana, 2016). It is evident that school closures during the coronavirus pandemic altered the daily lives of the students and their families and how children around the world were forced to become virtual-school learners while their parents had to assume the role of pseudo-teachers (Cohen & Kupferschmidt, 2020; Daniela et al., 2021; Garbe et al., 2020). These closures are likely to have damaged children's psychological and educational development and have caused loss of income and productivity in adults (Edmunds, 2020; Macartney et al.,2020). Millions of students were exposed to life changing impacts on their living environments, daily routines, and educational and social-relational networks: critical contexts that promote mental health and resilience to traumatic events (Dalton et al., 2020; Danese et al., 2020). Considering the vital importance of routine school settings for the healthy development of children (Catalano et al., 2004; Segre et al., 2021) and the evidence that education "is health" (UNESCO, 2016), it is regrettably imaginable how this situation could have an amplified impact on children with sensory disabilities. In fact, previous studies have shown that deafness is already associated with significant heterogeneity in cognitive, social and emotional development (Holt et al., 2020; Kral & O'Donoghue, 2010). However, the psychological studies related to SARS Cov 2 and SARS Cov 1 primarily focused on the psychological status of typical people during the pandemic (Yang et al., 2021) and rarely, if at all, focused on deaf students (i.e., Alqraini & Alasim, 2021).

Studies have investigated the stress that parents experience associated with their HL children (Blank et al., 2020; Quittner et al., 2010), who may also face many other difficulties during distance learning courses because of the barriers their children face (Mantzikos & Lappa, 2020; McKeown & McKeown, 2019). Furthermore, a recent study conducted at the beginning of the coronavirus pandemic has highlighted the difficulties faced by the parents of children with special needs (SNs) who exert more effort when taking care of their children's learning and living conditions than that experienced by parents of typically developing children (Ren et al., 2020).

1.1 | The current study

The primary aim of this study, titled COCLOVID (evaluation of the online **Di**dactic in deaf children with or without **COChLe**ar implants and on their parents during the **COVID**-19 pandemic), was to investigate the psychological characteristics of online learning during the Covid-19 lockdown on Italian students with and without hearing impairments and on their parents who were forced to isolate and undertake social distancing. In particular, we first investigated the possible influence of auditory features on Anxiety levels and School Wellbeing in the participants to try to answer the following research questions:

(1) Does having hearing difficulties or being the parent of such a child impact on School Wellbeing experienced during the period of online education?
(2) In the light of recent studies concerning the psychological and social effects of the Covid-19 pandemic on young and adult populations, are there any differences in the psychological costs between children with or without hearing impairments and on the parents of healthy or hearing-impaired children?
(3) Are there any significant correlations between psychological experiences and perceptions associated with School Wellbeing amongst students and their parents during the quarantine period?

To our knowledge, the experimental evaluation of School Wellbeing and Anxiety during online learning due to the Covid-19 pandemic has received very little attention, especially concerning students with hearing impairments and the impacts upon their families. To date, we are not aware of any other study that has drawn attention to this specific topic.

2 | METHODS

2.1 | Study design and participants

COCLOVID is a study based on disseminating an ad hoc prepared online survey, managed through EUSurvey, a web platform promoted by the European Commission (2013). Questions were designed to collect and highlight the socio-demographic data of parents and their children and on their opinions about school closures and online learning; the delivery of a standardized questionnaire enabled the assessment of the psychological dimensions of participants. The survey was broadcast through (i) email invitations to personal contacts and via healthcare professionals and their patients, (ii) social media channels. The survey was available from May to August 2020, taking approximately 30 min to complete. Participants were informed about the aims of the study and, before starting, electronic informed consent was requested from each parent and child. Participation in the study was voluntary, and therefore participants did not receive compensation for taking part in it. Data collection was conducted according to the principles outlined in the Helsinki Declaration of 1975, revised in 2000, and was approved by the Institutional Ethics Committee of Policlinico Umberto I, Rome, Italy (No. 259/2020).

2.2 | Outcomes

2.2.1 | General information

The survey questions collected socio-demographic information (age, gender, education, occupation, region of origin, deafness, use of hearing aids or devices) and opinions about online learning.

2.2.2 | School wellbeing

The "Questionnaire on School Wellbeing" (QBS; Tobia & Marzocchi, 2015a), assessed children's (aged 8–13) wellbeing at school. It is based on a multidimensional concept of School Wellbeing that includes psychological, cognitive, and social components using a three-perspective approach (indeed, the questionnaire investigates student, parent and teacher observations). In this study, we considered School Wellbeing from the perspective of students and parents.

The student version consists of 27 items and investigates the subjective school experience of male and female students attending primary school (3rd to 5th grade) and middle school (6th to 8th grade) by examining 5 QBS scale scores: (1). *Gratification obtained by school results*—GBS; (2). *Relationship with classmates*—RWC; (3). *Relationship with teachers*—RWT; (4). *Emotional attitude towards school*—EA; (5). *Self-efficacy*—SE and finally, a Total School Wellbeing—TOT score was obtained by combining the QBS scales scores. The parental version comprised of 36 items and five scales: (1). *Personal experience in relation to the child's difficulties*—PE; (2). *Evaluation of learning processes*—ELP; (3). *Child's emotional difficulties at school*—ED; (4). *Child's awareness of his/her difficulties*—CA; (5). *Relationship with teachers*—RWT.

With respect to the objectives of this study, only scales ELP, ED, RWT have been considered for parents. The "Multidimensional Self-Esteem Test" (TMA, Bracken, 1993) is based on a hierarchical model of self-esteem: it is comprised of six self-esteem dimensions (*Personal*, *Skills*, *Emotional*, *School*, *Family*, and *Body*); the measure also includes a scale testing Total self-esteem. The test consists of six groups of 25 items for each dimension explored, and each item requires one of 4 possible answers: absolutely true, true, not true, absolutely not true. The test provides scores on six rating scales corresponding to the six self-esteem dimensions, and the Total self-esteem-related scores. The average scores for self-esteem in the normative sample are between 85 and 115. In line with the aims of the study, we administered School and Family dimensions for 14–19-year-old students. The latter scales were related to the "General Self-Efficacy scale" (GSE, Schwarzer & Jerusalem, 1995) to evaluate the individual's Self-Efficacy. GSE is comprised of 10 items, scored on a 4-point scale from 1 (*not at all true*) to 4 (exactly true); higher values indicate higher self-efficacy. GSE psychometric characteristics have been extensively studied across several countries by Scholz et al. (2002), the Italian version (Sibilia et al., 1995) was used.

2.3 | Anxiety

The "State-Trait Anxiety Inventory for adults" (STAI-Y, Spielberger et al., 1983; for Italian adaptation, see Pedrabissi & Santinello, 1989) was used with parents. It is comprised of separate self-reporting scales for measuring *State* (S) and *Trait* anxiety. The S-Anxiety scale (STAI Form Y-1) consists of 20 statements that evaluate how respondents feel "right now, at this moment." The T-Anxiety scale (STAI Form Y-2) consists of 20 statements that assess how people generally feel. Participants answered 40 items on a 4-point Likert scale ranging from 0 (*not at all*) to 4 (*very much so*). The range score for each scale is 20–80, with the higher scores indicating greater anxiety.

The "Revised Children's Manifest Scale" (RCMAS-2; Reynolds & Richmond, 1985; for Italian adaptation, see Reynolds et al., 2012) was used with students. RCMAS-2 assesses both the degree and quality of anxiety experienced by children and adolescents (aged 6–19). It is a relatively brief instrument (49 items) suitable for administration in both clinical and educational settings. It is one of the most widely used questionnaires employed when researching and treating developmental anxiety (Gerard & Reynolds, 2014). Scores are provided for five distinct scales: *Physiological Anxiety* (PHY), *Concern* (CON), *Social Anxiety* (SOC), *Total Anxiety* (TOT), *Defensive Attitude* (DEF).

2.4 | Statistical analysis

Descriptive statistics were performed to describe the sociodemographic and hearing characteristics, as well as opinions about online-learning during Covid-19 related aspects, in the parent and child populations. After checking the normality of each data distribution with both Shapiro–Wilk and Kolmogorov–Smirnov tests, independent *t* tests were used to compare the effects of Independent Variables (Auditory Condition [Normal Hearing {NH}/HL]; Gender [Male/Female]; School Grade [Elementary/Middle]) on Dependent Variables (TOTAL School Wellbeing; TOTAL School Self Esteem; TOTAL Family Self-Esteem; TOTAL Self Efficacy and TOTAL Anxiety) in student groups and Trait

Anxiety and State Anxiety, in parent groups. Subsequently, Factorial analysis of variance (ANOVAs) were performed for each questionnaire to investigate the influence of Independent Variables on the considered subscales. Independent Variables were: Auditory Condition (NH/HL), Hearing Groups (Unilateral cochlear implant user-UCI, Bilateral cochlear implant user-BCI, Bimodal hearing device user-BIM, Hearing aid user-HA, Normal hearing-NH), Gender (M/F), School Grade (Primary/Middle), Parent Education (4 levels) and Family Income (4 levels); while the investigated questionnaire scales considered as Dependent variables were: QBS student scales (5 levels: GBS/RWC/RWT/EA/SE), QBS parent scales (3 levels: ELP/ED/RW) RCMAS-2 scales (4 levels: PHY/CON/SOC/DIF), TMA scales (2 levels: SCH/FAM); STAI scales (State/Trait). Duncan's post hoc test (Duncan, 1955) was used to investigate statistically significant results of ANOVA tests; partial eta squared (η_p^2) (Cohen, 1973, 1988) were computed as measures of the effect size for each dependent variable. Finally, Pearson's Correlation Analysis (r) was performed to explore the correlation between study variables; p values of less than 0.05 were considered statistically significant.

3 | RESULTS

3.1 | Demographic characteristics

The characteristics of all participants are shown in Table S1. The survey was fully completed by 65 pairs (parent & child-student) of participants. Due to the aims of the present research, the inclusion criteria adopted were for children aged between 8 and 19 years of age and who had no concurrent neuropsychiatric disorders; four pairs were therefore excluded from the study for declaration of diagnosed neuropsychiatric disorders. The final experimental population was composed of 51 mothers (83.607%, mean age 44.09 ± 6.23), 10 fathers (16.393%, mean age 49.66 ± 4.69), 25 sons (40.98%, mean age 12.33 ± 3.01) and 36 daughters (59.02%, mean age 12.19 ± 2.96). Amongst these 61 children, 43 of them were Hearing Device users divided into Unilateral Cochlear Implant users (UCI), Bilateral Cochlear Implant users (BCI), Bimodal Hearing Device users (BIM) and Hearing Aid (HA) users. Two student groups were created: (i) from 3rd to 8th grade = Junior Student group—JS ($N = 45$, mean age = 11 ± 1.73); (ii) attending high school = Senior Student group—SS ($N = 17$, mean age = 16.11 ± 1.21). According to hearing aid characteristics, in the JS group respectively, there were 7-UCI, 10-BCI, 8-HA, 9-BIM, and 11 NH; in the SS group, there were 6-UCI, 1-BCI, 2-HA, and 7-NH. Educational levels showed 9 (14.754%) parents with a Secondary School Diploma, 22 (36.066%) with a High School Diploma (13 years of study), 24 (39.344%) with a Bachelor's or Master's Degree, and 6 (9.836%) with a Post-Graduate Degree. According to the responses to the questionnaire regarding online learning, overall parents (45.902%) reported a "*quiet*" level of concern about school closures with 4 parents of HL and 2 of NH children professed to be "*extremely*" concerned. For 54.098% of parents, online learning is "*quite useful*" whereas for 8 parents of HL children, compared to no parents of NH children, found it to be "*very useful*". Overall students (49.180%) reported it to be "*quite*" enjoyable taking lessons online with 4 HL students, compared to no NH students, considering it to be "*extremely*" enjoyable taking online classes. The overall opinion about online lessons is that they were "*normal*" for 45.902% of students while 8 HL and 6 NH students considered it to be "*tiring*." Only HL students (18.605%) described them as "*difficult to follow*."

3.2 | The general impact of conditions (NH/HL, M/F, middle/elementary) on anxiety and school wellbeing

The results of the t test conducted to compare Auditory Condition, Gender, and School Level variables are presented in Table 1. Considering all psychological assessments, there were no statistically significant differences between student and parent groups concerning all conditions ($p > 0.05$).

TABLE 1 Results of *t* test for independent sample by groups

(A)	µ HL group	µ NH group	*t* value	*df*	*p*	Valid n HL	Valid n NH	*SD* HL	*SD* NH
TOTAL School Wellbeing QBS (JS)	47	52.272	−1.299	43	0.200	34	11	11.88	11.055
TOTAL School self-esteem scale—TMA (SS)	108.555	107.571	0.303	14	0.766	7	9	3.954	8.502
TOTAL Family self-esteem scale—TMA(SS)	107.333	109.571	0.395	14	0.693	9	7	10.723	11.900
TOTAL Self-efficacy scale (SS)	60.449	58.689	−1.000	14	0.189	9	7	2.902	1.928
TOTAL Anxiety RCMAS-2 (all students)	48.323	43.222	−1.575	59	0.120	43	18	10.487	13.171
State Anxiety—STAI-Y Test (Parents)	44.046	44.444	0.146	59	0.884	43	18	10.980	5.305
Trait Anxiety—STAI-Y Test (Parents)	40.209	38.055	−0.933	59	0.354	43	18	8.730	6.786
(B)	**µ F group**	**µ M group**	***t* value**	***df***	***p***	**Valid *n* F**	**Valid *n* M**	***SD* F**	***SD* M**
TOTAL School Wellbeing QBS (JS)	50.444	45.055	1.525	43	0.134	27	18	12.911	9.276
TOTAL School self-esteem TMA (SS)	109.000	107.000	0.623	14	0.543	9	7	5.612	7.257
TOTAL Family self-esteem TMA (SS)	102.333	106.000	−0.380	14	0.709	9	7	23.590	10.503
TOTAL Self-efficacy scale (SS)	39.888	41.285	−0.495	14	0.628	9	7	4.106	7.111
TOTAL Anxiety RCMAS-2 (all students)	47.388	45.840	0.515	59	0.607	36	25	12.504	9.956
State Anxiety—STAI-Y Test (Parents)	45.388	42.400	1.198	59	0.235	36	25	10.232	8.534
Trait Anxiety—STAI-Y Test (Parents)	40.611	38.80	1.188	59	0.239	36	25	8.445	7.777
(C)	**µ Primary Students**	**µ Middle Students**	***t* value**	***df***	***p***	**Valid *n* primary students**	**Valid *n* middle students**	***SD* primary students**	***SD* middle students**
TOTAL School Wellbeing QBS (JS)	46.190	50.125	1.120	43	0.268	21	24	11.877	11.604
TOTAL Anxiety RCMS-2 (JS)	48.190	46.375	−0.484	43	0.630	21	24	11.298	13.509
State Anxiety—STAI-Y Test (Parents of JS)	45.333	45.375	0.013	43	0.988	21	24	10.233	9.863
Trait Anxiety—STAI-Y Test (Parents of JS)	41.476	39.166	−0.941	43	0.351	21	24	8.394	8.052

Note: The table shows no differences between groups (A:HL/NH, B:M/F, C:Middle/Elementary) on Dependent variables (School Wellbeing, Anxiety).

Abbreviations: F, female; HL, hearing loss; M, male; NH, normal hearing; QBS, Questionnaire on School Wellbeing; *SD*, standard deviation.

3.3 | School wellbeing

In the JS group, ANOVA results did not show an effect of the Auditory Condition variable on QBS scales ($F_{(1,43)} = 0.992$, $p = 0.324$, $\eta_p^2 = 0.022$), but did show a significant difference within QBS scales ($F_{(4,172)} = 3.432$, $p = 0.009 = 0.073$). Post hoc analysis showed higher GBS and EA scores recorded rather than for the other three QBS scales (Figure 1).

With regard to QBS scales, a nonsignificant effect within the variable Hearing Groups was found ($F_{(4,40)} = 2.226$, $p = 0.083$, $\eta_p^2 = 0.182$); statistically significant differences amongst the QBS scales ($F_{(4,160)} = 8.720$, $p < 0.001$, $\eta_p^2 = 0.178$) (Figure 1) and a significant interaction between QBS scales and Hearing Groups ($F_{(16,160)} = 2.352$, $p = 0.003$, $\eta_p^2 = 0.190$) were found. Post hoc analysis showed differences between QBS scales similar to the ones found in Figure 1 and nonhomogeneity of QBS scores considering the effect of auditory characteristics (Figure 2). Several significant differences between and within Hearing Groups emerged from the analysis (Table S3). First, we observed numerous differences in QBS values between and within hearing loss groups (UCI, BCI, HA, BIM) but only slight differences between NH and HL groups and a total absence of differences within the NH group. A general observation concerning comparison amongst groups is that the HA group reported more statistically significant differences in the pairwise comparisons than with other Hearing Groups' QBS subscales scores.

QBS results in the parent group (of junior students), did not show any effect ($F_{(4,40)} = 0.859$, $p = 0.496$, $\eta_p^2 = 0.079$), while there was a significant difference amongst QBS scales ($F_{(2,86)} = 17.240$, $p < 0.001$, $\eta_p^2 = 0.286$), respectively. Post hoc analysis showed lower scores for the RWT scale than for ELP ($p \leq 0.001$) and ED ($p \leq 0.001$) (Figure 3). Moreover, QBS scales were not influenced by either Gender (of the child) ($F_{(1,43)} = 0.042$, $p = 0.839$, $\eta_p^2 = 0.000$) or Income ($F_{(3,41)} = 0.736$, $p = 0.536$, $\eta_p^2 = 0.051$).

We observed, however, a significant effect of parents' Education Level on QBS scales ($F_{(6,82)} = 2.626$, $p = 0.022$, $\eta_p^2 = 0.161$). Post hoc analysis showed that the only significant difference in the same scale between education levels emerged for RWT (i.e., the QBS scale that assesses the relationship and level of trust that parents have with their child's teachers): the values of parents with lower education levels were significantly higher in comparison to parents with High School ($p = 0.005$), Graduate ($p = 0.003$) and Postgraduate ($p = 0.011$) levels, respectively (Figure 4).

The results of ANOVA for the SS group showed that TMA scores were not influenced either by the factor Gender ($F_{(1,12)} = 0.000$, $p = 0.992$, $\eta_p^2 = 0.000$) or by the factor Hearing ($F_{(1,12)} = 0.5079$, $p = 0.489$, $\eta_p^2 = 0.040$).

3.4 | Anxiety

In all student participants, the overall results of factorial ANOVA analysis did not show any effect of either Auditory Condition ($F_{(1,57)} = 0.243$, $p = 0.623$, $\eta_p^2 = 0.004$), or Gender ($F_{(1,57)} = 0.182$, $p = 0.670$, $\eta_p^2 = 0.003$), or Hearing Groups ($F_{(4,56)} = 0.416$, $p = 0.795$, $\eta_p^2 = 0.028$) whereas a strong significant difference was observed amongst RCMAS-2 scales ($F_{(3,171)} = 7.897$, $p \leq 0.001$, $\eta_p^2 = 0.121$). The post hoc analysis showed significantly higher mean scores for the subscale *Defensive Attitude* than the other three scales (Figure 5).

In the Parent group, the results for the STAI-Y scales × Auditory Condition × Gender factorial ANOVA analysis did not show any impact on Auditory Condition ($F_{(1.57)} = 2.514$, $p = 0.118$, $\eta_p^2 = 0.042$] nor Gender ($F_{(1,57)} = 0.449$, $p = 0.505$, $\eta_p^2 = 0.007$) on parent's anxiety levels but revealed significant differences between STAI-Y scales ($F_{(1.57)} = 25.929$, $p = 0.000$, $\eta_p^2 = 0.312$]. Post hoc analysis showed higher scores on State than on the Trait scale ($p \leq 0.001$) (Figure 6).

In addition, further investigation into the effects of variables on anxiety levels did not show any significant variations for the parent Education Level ($F_{(3,57)} = 2.078$, $p = 0.113$, $\eta_p^2 = 0.098$) nor for the family Income level ($F_{(3,57)} = 0.468$, $p = 0.705$, $\eta_p^2 = 0.024$).

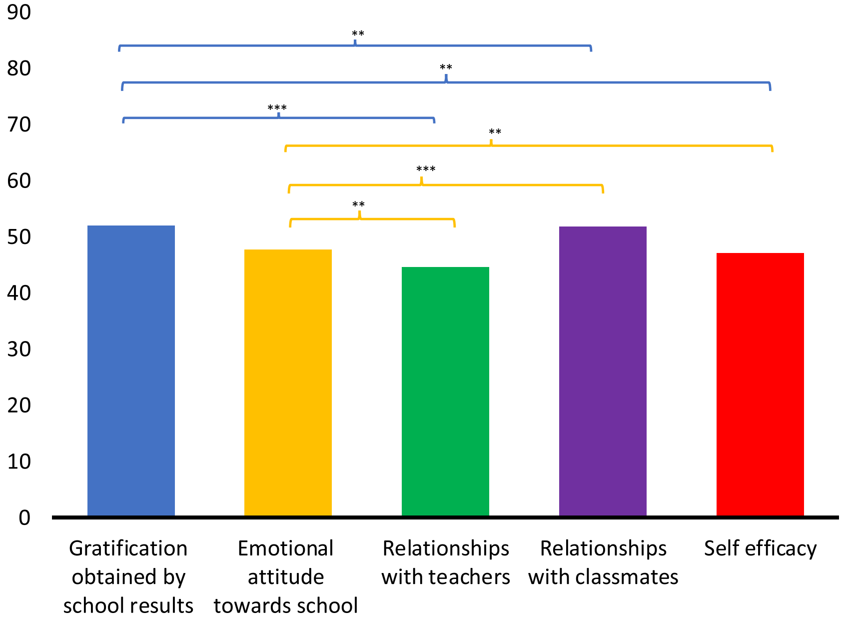

FIGURE 1 Junior Students School Wellbeing. The graph evidences the differences between QBS scales. Significant differences between QBS scales emerging from post hoc Duncan's test are indicated (*$p \leq 0.05$; **$p \leq 0.01$; ***$p \leq 0.001$). Bars describe means, and error bars describe standard deviations

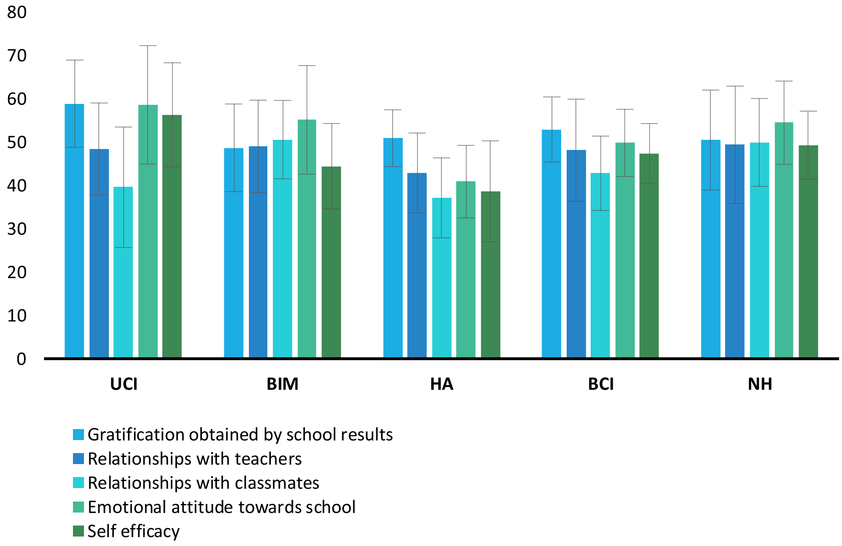

FIGURE 2 School wellbeing in Junior Students Hearing Groups. The graph shows the QBS scales values for JS Hearing Groups (for ANOVA's QBS scales × Groups post hoc analyses see Table S3). Bars describe means, and error bars describe standard deviations. ANOVA, analysis of variance; QBS, Questionnaire on School Wellbeing.

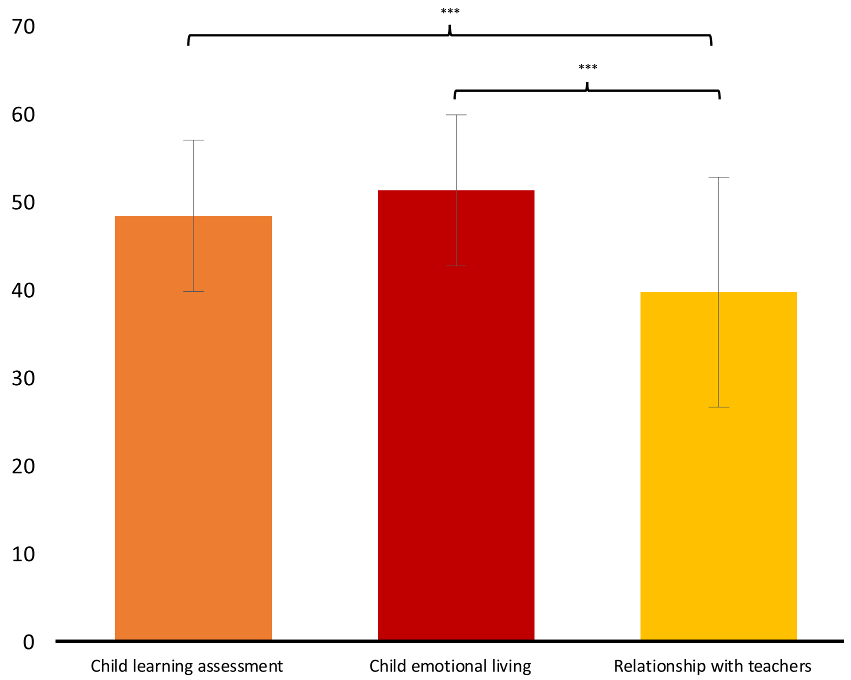

FIGURE 3 Parent's School Wellbeing. The graph shows significant differences between QBS parent's scales emerging from Duncan's Post hoc test (*$p \le 0.05$; **$p \le 0.01$; ***$p \le 0.001$). Bars describe means, and error bars describe standard deviations. QBS, Questionnaire on School Wellbeing.

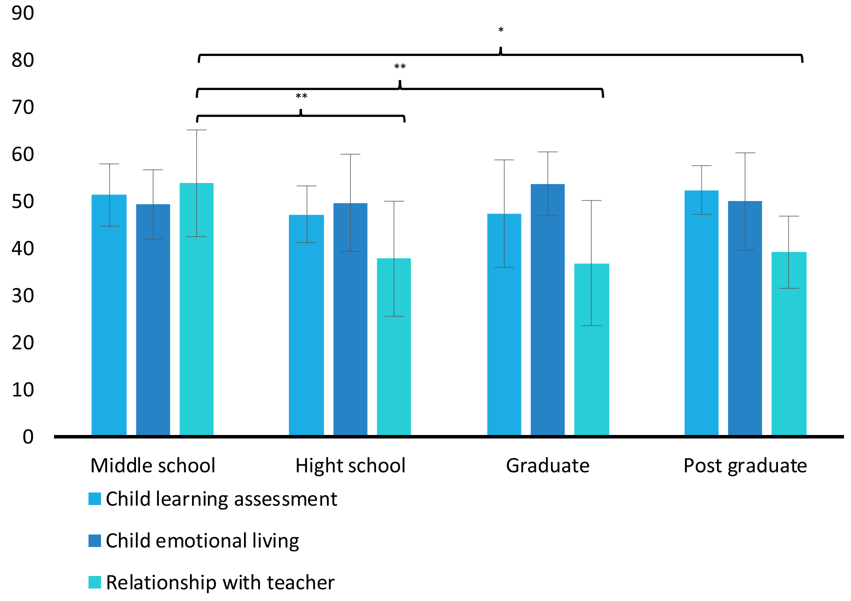

FIGURE 4 Parent's School Wellbeing for Education Level. The graph shows significant differences between QBS parent's scales and Education Level emerging from post hoc Duncan's test (*$p \le 0.05$; **$p \le 0.01$; ***$p \le 0.001$). Bars describe means, and error bars describe standard deviations. QBS, Questionnaire on School Wellbeing.

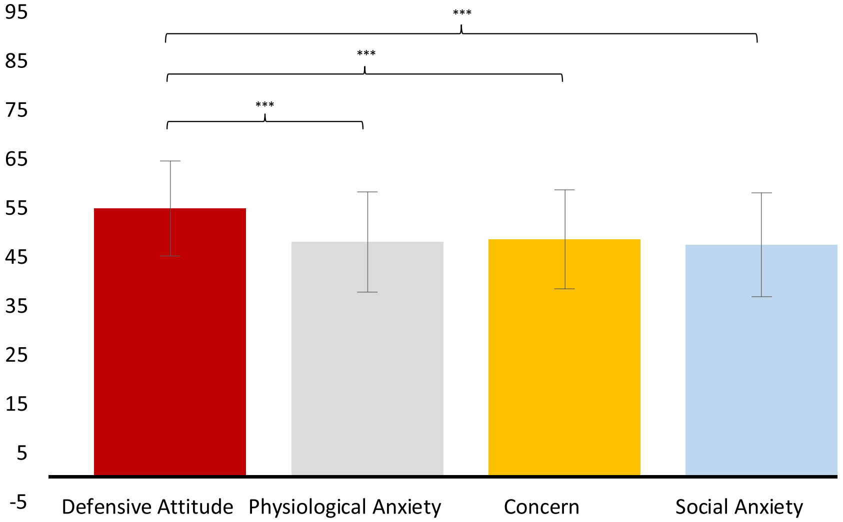

FIGURE 5 Anxiety in Student Group. The graph shows the significant differences between RCMAS-2 scales emerging from Post hoc Duncan's test (*$p \leq 0.05$; **$p \leq 0.01$; ***$p \leq 0.001$). Bars describe means, and error bars describe standard deviations

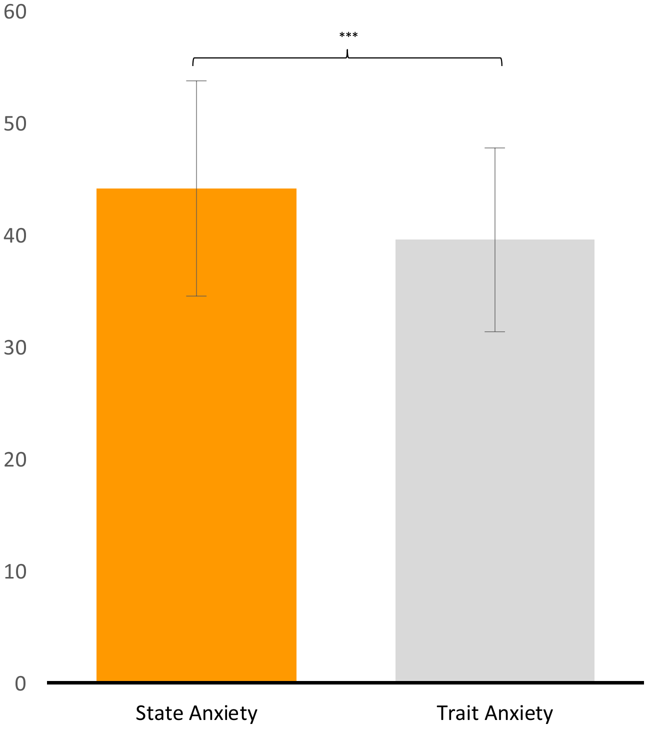

FIGURE 6 Anxiety in Parent Group. The graph shows the differences between State and Trait Anxiety STAI-Y scales. Post hoc Duncan's test indicated (*$p \leq 0.05$; **$p \leq 0.01$; ***$p \leq 0.001$). Bars describe means, and error bars describe standard deviations

3.5 | Factors correlated with anxiety, school wellbeing

The Pearson *r* coefficient was calculated between QBS scales and RCMAS-2 scales to investigate the relationship between School Wellbeing and Anxiety variables amongst TOTAL, NH, and HL in the JS group (Table 2). The results showed no significant values between QBS scales and RCMAS-2 scales in the Total (45) JS sample. However, considering HL and NH separately, results showed that in the HL group a negative correlation between *Defensive Attitude* and RWT ($r = -0.35$, $p < 0.05$) was absent in NH group. In the latter group strong positive correlations were noted between *Defensive Attitude* and RWC ($r = 0.85$, $p < 0.05$) and between *Defensive Attitude* and SE ($r = 0.72$, $p < 0.05$). In the Parent group the results of the correlation analysis between the answers to qualitative items within the survey and, respectively, QBS (parent) scales and STAI-Y scales are shown in Table 3 **(A** and **B)**. As inferred from the results, there was a strong negative correlation between State anxiety and the level of education of the NH parent group ($r = -0.66$, $p < 0.05$) (Table 3 [**B**]). Moreover, we observed a relationship between overall Parent and Student anxiety variables ($0.34 \le r \le 0.64$, $p < 0.05$), as shown in Table 4.

4 | DISCUSSION

4.1 | School wellbeing

4.1.1 | Students

Overall, the student group presents values on QBS scales in the average when compared to the reference norms (Tobia & Marzocchi, 2015a) (Figure 1). The direct comparison of NH and HL Young Students on the TOT-QBS scale does not reveal significant differences (Table 1 [**A**]): the deafness factor does not affect the Total School Wellbeing results in the studied sample. This outcome may not be in line with research showing that deaf children experience disadvantages in the educational system (Berry, 2017). However, it is in line with studies showing that most cochlear implanted children in mainstream schools seem to have a positive attitude towards self-esteem and confidence (Choi et al., 2016). Moreover, as Rotsika et al. (2011) suggested, struggling children (like deaf children) may underestimate their problems to protect themselves from the pain of facing their difficulties. This assumption could be supported by the fact that compared to their NH peers, of whom only 5.556% consider online lessons "*funny*," 0.00% to be "*difficult*" and 16.667% "interesting." Whereas for HL students, online classes are considered by 11.628% to be "*funny*," 18.605% to be "*difficult to follow*" but for 18.605% of them to be "*interesting*" (Table S1).

Even considering the five QBS scales, we have seen that no significant differences between NH/HL children emerged. Ayfer and Ocakçi (2012) found that children with HL, compared to NH using the Kid_KINDL scale for the evaluation of quality of life (for details, see Lin et al., 2014), had significantly lower scores in terms of emotional, family, School Wellbeing and self-esteem results in the scales of the questionnaire. Furthermore, another study (Yigider et al., 2020) reported that the leading quality of life scores of children with HL, according to the same Kid- KINDL scale, were significantly lower than for healthy children. However, the Authors did not find differences between NH and HL groups in terms of family and school scales, but the HL group had significantly lower scores in emotional wellbeing and social relationships (psychological variables also present in QBS). Therefore, the above-cited literature shows that hearing loss may dramatically reduce quality of life in the pediatric population. These observations give rise to further important considerations.

Self-Esteem is a crucial component in human beings' psychological wellbeing and life satisfaction (Borowiec et al., 2019; Rosenberg, 1965). In addition, Self-Esteem, as a component of emotional wellbeing is one of the dimensions of quality of life (Knox & Muros, 2017). Moreover, a global report on disability (World Health Organization-WHO, 2011) highlighted the need to undertake research and make interventions to improve quality of life and its dimensions among people with disabilities. In this perspective, studies concerning the Self-Esteem of

TABLE 2 Relationships between school wellbeing (QBS) and anxiety (RCMAS-2) in total student population

RCMAS-2	Defensive Attitude			Total Anxiety			Physiological Anxiety			Concern			Social Anxiety		
QBS scales (Children)	TOT JS	NH JS	HL JS	TOT JS	NH JS	HL JS	TOT JS	NH JS	HL JS	TOT JS	NH JS	HL JS	TOT JS	NH JS	HL JS
GBS	−0.09	0.1	−0.14	0.03	0.35	−0.18	−0.13	−0.24	−0.12	−0.26	−0.42	−0.22	−0.12	−0.12	−0.13
RWT	−0.23	0.11	**−0.35***	0.14	0.29	0.1	0.05	0.11	0.04	0.1	−0.03	0.16	0.07	0.09	0.07
RWC	0.14	**0.85***	−0.02	−0.05	−0.15	0.09	0.05	0.18	0.06	0.08	0.11	0.09	0.16	0.57	0.07
EA	0.19	0.37	0.15	−0.18	−0.33	−0.1	−0.08	0.01	−0.08	−0.08	0.15	−0.12	0.01	0.34	−0.07
SE	0.22	**0.72***	0.13	−0.16	−0.99	−0.15	−0.03	0.26	−0.07	−0.18	−0.14	−0.18	−0.07	0.2	−0.13
TSW	0.04	0.42	−0.05	−0.12	−0.1	−0.08	−0.08	−0.03	−0.07	−0.12	−0.21	−0.08	0	0.2	−0.05

Note: In bold, * = correlation (*r*) significant at $p < 0.05$.

Abbreviations: EA, emotional attitude towards school; GBS, gratification obtained by school results; QBS, Questionnaire on School Wellbeing; RWC, relationship with classmates; RWT, relationship with teachers; SE, self efficacy; TSW, total school wellbeing.

TABLE 3 Relationships between Qualitative Items and School Wellbeing (QBS) and Anxiety (STAY-Y) in Parents (P) of all Students and Junior Students (JS).

Items		*Income*			*Instruction*			*Internet connection at home*			*Opinion about online learning*			*Is your child wasting time?*	
5 A QBS scales[a]	P_TOT JS	P_NH _JS	P_HL _JS	P_TOT_ JS	P_NH_ JS	P_HL_ JS	P_TOT_ JS	P_NH_ JS	P_HL_ JS	P_TOT_ JS	P_NH _JS	P_HL _JS	P_TOT_ JS	P_NH_JS	P_HL _JS	P_TOT_ JS
ELP	0.2 4	0.38	0.25	0.01	0.08	0.02	0	0.01	0.01	0.21	0.29	0.19	0.2	0.17	0.24	−0.12
ED	0.1 5	0.27	0.25*	0.13	−0.26	0.17	−0.06	0.08	−0.1	0.13	0	0.16	0.03	−0.51	0.14	−0.21
RWT	1	0.06	0.02	−0.28	−0.15	−0.3	0.05	−0.36	0.17	0.27	0.21	0.29	0.17	0.74*	0.04	−0.07
5 B STAI-Y	P_TOT	P_NH	P_HL	P_TOT	P_NH	P_HL	P_TOT	P_NH	P_HL	P_TOT	P_NH	P_HL	P_TOT	P_NH	P_HL	P_TOT
Sate	0.0 2	−0.1	0.04	0.09	− 0.66*	0.20	−0.01	0.54*	0.11	−0.27*	−0.3	− 0.28	−0.09	0.07	−0.13	0.25
Trait	0.0 6	0.001	0.17	0.06	−0.45	0.21	−0.25	0.59*	−0.11	−0.16	−0.12	− 0.18	−0.04	0.24	−0.11	0.16

Items	*Is it beneficial to take classes online?*			*Presence of parent during online lessons*			*Need of Additional tools*			*Concern about school closure*		
5 A QBS scales[a]	P_TOT JS	P_NH _JS	P_HL_JS	P_TOT_ JS	P_NH_JS	P_HL_JS	P_TOT_ JS	P_NH_JS	P_HL_JS	P_TOT_JS	P_NH_JS	P_HL_JS
ELP	0.2 4	−0.14	−0.14	−0.39*	−0.28	0.54*	0	−0.57	0.28	−0.29	−0.31	−0.28
ED	0.1 5	−0.3	−0.17	−0.31*	−0.53	−0.25	0.15	−0.35	0.28	−0.13	−0.47	−0.1
RWT	1	0.4	−0.19	−0.02	0.5	−0.13	−0.08	0.2	−0.15	−0.21	−0.17	−0.21
5 B STAI-Y	P_TOT	P_NH	P_ HL	P_TOT	P_NH	P_HL	P_TOT	P_NH	P_HL	P_TOT	P_NH	P_HL
Sate	0.0 2	0.38	0.38	0.34*	0.12	0.38*	−0.02	−0.1	−0.19	0.38	−0.54*	0.4*
Trait	0.0 6	0.37	0.2	0.21	0.11	0.2	−0.06	−0.23	−0.2	0.25	−0.59*	0.23

In red

*=correlation (r) is significant at $p < 0.05$ level.

[a]=abbreviations of QBS parent's scales:

ELP-Evaluation of learning processes; ED-Child's emotional difficulties at school; RWT-Relationship with teachers.

TABLE 4 Relationships between anxiety in parent (STAI-Y) and anxiety in student (RCMAS-2)

RCMAS-2	**Defensive Attitude**			**Total Anxiety**			**Physiological Anxiety**			**Concern**			**Social Anxiety**		
STAI-Y	**TOT POP**	**TOT NH**	**TOT HL**	**TOT POP**	**TOT NH**	**TOT HL**	**TOT POP**	**TOT NH**	**TOT HL**	**TOT POP**	**TOT NH**	**TOT HL**	**TOT POP**	**TOT NH**	**TOT HL**
State Anxiety	−0.03	**0.58***	−0.13	**0.34***	0.41	**0.37***	**0.43***	**0.60***	**0.41***	**0.35***	**0.67***	**0.31***	**0.30***	**0.54***	0.26
Trait Anxiety	−0.04	0.45	−0.18	**0.37***	0.39	**0.35***	**0.39***	0.39	**0.39***	**0.46***	**0.65***	**0.39***	0.25	**0.58***	0.21

Note: In bold, * = correlation (*r*) significant at $p < 0.05$ level.

HL people observed a lower Self-Esteem level among HL children compared with their peers (Borowiec et al., 2019; Lesar & Smrtnik Vituli, 2014). Studies also show that the leading causes of lower Self-Esteem in HL children are difficulties in communication and the inability to form peer relationships (Fellinger et al., 2012). Moreover, Self-Efficacy is predictive of higher Self-Esteem (Stroiney Hermann, 2005) and has been applied to such diverse areas as school achievement and emotional disorders (Schwarzer et al., 1997). According to Bandura (1997), Self-Efficacy makes a difference in how people feel, think and act: low Self-Efficacy is associated with low Self-Esteem: both Self-Esteem and Self-Efficacy, when measured, showed significant positive intercorrelations (Lane et al., 2004). Our results do not show differences between NH and HL children in QBS SE. However, in the light of the literature, we should have expected significantly lower scores in HL students on the QBS scales, specifically concerning SE, EA, and GBS scales. To explain this lack of difference between HL and NH, we can advance two hypotheses:

(i). During lockdown, the family environment has been a strong protective factor for School Wellbeing perceived by the HL child allowing this group to have QBS scores not significantly different from the NH group. This hypothesis could be verified by repeating the study after the return to a face-to-face didactic setting.

(ii). Given the different auditory features in HL students, it could be assumed that the nonhomogeneity of deafness characteristics in the HL group could increase their overall mean scores resulting in the loss of any differences when compared to the NH group. Whilst exploring this aspect, we performed a further analysis of the Hearing Groups variable (Figure 2). Results revealed significant differences between students with different hearing devices: the characteristics of deafness in the HL groups could significantly increase the mean scores of School Wellbeing. Such evidence suggests the need for further investigation and is probably explained by a high heterogeneity due to the HA clinical condition. More specifically, the evidence of a nonhomogeneity of the QBS scales' scores both *within* and *between* HL subgroups and the homogeneity found *within* the NH group (Table S3), could be one of the causes of the lack of significant differences in QBS scores between NH and HL. Based on this observation, analysis of future data possibly taken in a regular didactic setting could refine and validate whether the audiological characteristics of HL students could play a decisive role in this lack of statistical variation.

A variable that reportedly influences not only School Wellbeing but also its potential predictors is Gender (Løhre et al., 2010) and consequently we investigated the impact of the Gender variable on TOTAL School Wellbeing. The main finding of an interesting study was that girls within each school level rated School Wellbeing more positively than boys even though the latter had fewer symptoms than girls (Konu & Lintonen, 2006). Similarly, Hascher and Hagenauer (2011) reported that female students had a more positive attitude towards school and greater enjoyment of it, but that they also had more somatic complaints and worries than male students. The latter were more distressed than females, but no significant gender effect was observed on depressive symptoms (Correia & Dalbert, 2007; Peter et al., 2013). In contrast, it has been reported that girls generally have higher negative affect scores and lower positive affect scores than boys (Clark & Watson, 1991), but at the same time, girls are more motivated to study than boys (Alivernini et al., 2018; Grouzet et al., 2006). Others assert that gender is a relevant factor in students' feelings at school: being female negatively affects it (Alivernini et al., 2018). Our initial analysis to assess whether Gender influences the Total School Wellbeing variable does not reveal any significant difference between M and F groups (Table 1 [**B**]). Although this result may be at odds with parts of the literature, one might speculate, as proposed by Savoye et al. (2015), that other factors related to wellbeing could interact with gender differences in School Wellbeing within our junior student sample.

We could suppose that during the pandemic period characterized by online teaching, neither the Gender factor nor Hearing (NH/HL) influenced the School Wellbeing of the students neither globally (TOT-QBS) nor at the level of the internal components (QBS scales). This observation can be considered partially in accord with the study on School Wellbeing in children with special educational needs-SEN (Tobia & Marzocchi, 2015b) that highlighted higher scores for control subjects in the scales of GBS, RWT, and SE, while no significant differences emerged between RWC and EA. Other findings have shown that children with SEN tend to have lower subjective wellbeing levels than children without SEN when talking about their schools (Barnes & Harrison, 2017).

School wellbeing also appeared to be significantly different between schools and between classes in the same school (Holfve-Sabel, 2014). For example, a study found that students in 4th-6th grades experienced better school conditions, social relationships, and self-fulfillment than students in 7th−12th grades (Konu & Lintonen, 2006). Comparing Middle and Elementary students, our data did not show significant differences in Total School Wellbeing (Table 1 [**C**]). This result contrasts with a QBS study that supported a higher level of School Wellbeing for the younger students (Tobia et al., 2019) but is in line with a study that compared associations across all school grades (Løhre et al., 2010), where a substantial effect of scholar grade on School Wellbeing variables was not observed.

Moreover, the scales used to evaluate School Wellbeing in the senior student populations were not found to be influenced by either Auditory Condition or Gender factors. However, this lack of significant effect of the factor Gender on TMA-School and Family scales, is in line with the Italian validation of the TMA questionnaire, which shows significant differences for several scales but not for the two used in the present study (Bergamini & Pedrabissi, 2003).

4.1.2 | Parents

With regard to the QBS scales used to evaluate children's School Wellbeing as perceived by their parents, no significant differences emerged amongst parents of NH or HL children: the deafness factor does not seem to influence parents' assessment. Significant differences emerged in the Parent sample amongst the scales: the score on *Relationship with teachers* is significantly lower than scores on ELP and ED scales (Figure 3). These results resonate with the idea that for online-learning during Covid-19 quarantine, overall parents seem to have given lower ratings (but still in a medium average compared to the QBS reference norms in Tobia & Marzocchi, 2015a) to the relationship that their child had with their teacher than that of the assessment of learning and the emotional experience of the child. It may be that this difference is due to the lack of direct contact between students and teachers. Our results differ in part from those of a study by the same authors just mentioned (2015) that showed, in the comparison between SNs and healthy children and their parents, significant differences in all QBS parent's scales, except for the RWT scale. This latter result is in line with our study and seems to support the hypothesis that the parent's evaluation of the relationship with their teacher is not a variable related to the child's specific needs.

We did not find significant differences in the interaction between QBS parent scales and the Gender of the child. This result differs from data reported in Tobia & Marzocchi, 2015b; which observed that parents gave a more significant evaluation of School Wellbeing in the case of a daughter and evaluated them as more competent in learning processes and managing school material difficulties. However, statistical differences between the scales are not reported in the cited study, and therefore an evidence-based assessment cannot be made here. Our results, indeed, are not in line with the evidence indicating that females are more competent in school (e.g., Berchialla et al., 2011; Hyde & Linn, 2006; Pomerantz et al., 2002; Spelke, 2005) but may be partially ascribed to the current pandemic situation that could result in the reduction of the effect of the intervening variable Gender of the child in assessing the child's perceived academic well-being by the parent.

The Education Level of parents seems to influence the scores of the QBS scale (Figure 4). More specifically, the most substantial difference emerges for the RWT scale: parents with an inferior level of education (until middle school) reported higher scores on the evaluation of the teacher and so we can speculate that they were more involved in this relationship during the quarantine period. As evidenced by Mantovani and Gasperoni (2017), parental involvement varies due to various factors, which may also act ambivalently. For example, higher levels of education are positively associated with school participation (Crozier, 1999; Lee & Bowen, 2006; Peña, 2000;

Potvin et al., 1999); but, conversely, parents with college degrees may be less likely to participate due to lack of time (Bæck, 2010).

However, this consideration needs more data to come to an informed position which would deviate from the purpose of this study.

4.2 | Anxiety

Nowadays, anxiety disorders (American Psychiatric Association, 2013) are a major worldwide health problem with sizeable psychological, social, and economic costs (Beddington et al., 2008). During childhood and adolescence, anxiety phenomena are highly prevalent (e.g., Broeren & Muris, 2009; Craske et al., 2013), affecting around 10% of children and 20% of adolescents (Essau et al., 2012). They can negatively interfere with general wellbeing, social life, academic performance and development of social skills (Kessler et al., 1995; Pine et al., 1998), independently of culture (Crawford & Manassis, 2001). When compared to ordinary hearing people, children and adolescents with hearing impairment have been found to suffer more from behavioral and emotional problems, including social anxiety (Hindley, 2005). Recent studies on the Italian population indicated that the Covid-19 pandemic appears to be a risk factor for higher levels of anxiety in younger and older adults (e.g., Casagrande et al., 2020; Germani et al., 2020; Rossi et al., 2020). According to these studies, the assessment of the anxiety state of participants resulted in it being extremely important for a complete evaluation of School Wellbeing, especially in children with sensory disabilities.

4.3 | Students

Overall, the mean values for all types of anxiety investigated within the RCMAS-2 questionnaire amongst the entire student group and subdivisions in NH/HL, are "*not particularly problematic*" as reported in the Italian norms.

(Reynolds et al., 2012). Moreover, anxiety symptomatology does not differ significantly by hearing or gender of the student. (Table 1 [**A**]). Regarding deafness, our results would seem to be at odds with the literature regarding SNs students (e.g., learning disability-LD). In fact, children with LD have been found to show higher levels of trait anxiety (M. Bender, 1993; W. N. Bender & Wall, 1994) and a high rate of anxiety disorder (Beitchman et al., 1996); a meta-analysis confirms that students with LD experience higher levels of anxious symptomatology than do their non-LD peers (Nelson & Harwood, 2011). Furthermore, it is well known that auditory processing has a crucial role in language development (Moeller et al., 2007; Bailey & Snowling, 2002) and that school-aged children who are hearing impaired are five times more likely to suffer from emotional disturbance (Wolters et al., 2012). Even slight or mild hearing impairment can result in negative consequences in the psychological domain, and there is a significant relationship between delayed language, anxiety and emotional-related problems (Azab et al., 2015). In the light of the literature, we can assume that our sample has undifferentiated anxiety symptomatology based on difficulties related to deafness, and protective interventions towards children (HL) most at risk during the quarantine. This speculation may be further corroborated by comparison with the results of the data possibly collected during the return to face-to-face teaching.

The significantly higher score found for the *Defensive attitude* RCMAS-2 scale across all student participants compared to other scales (Figure 5) suggests that students are unwilling to admit common failings or have attempted to give a very positive self-image in a *naïve* or immature way. In fact, the *Defensive attitude* scale, containing items such as "I never get angry," "I like everyone I know," "I am always kind" is often used as an indicator of social desirability (Dadds et al., 1998) or/and defensiveness (Joiner et al., 1996). In some cases, high values express an excessive need for social desirability or acceptance (Reynolds & Richmond, 2008). As shown previously (Figure 5), *Defensive attitude* scale scores in our population are in a "not particularly problematic" range (see

Reynolds et al., 2012 for normative standard). However, the mean score is significantly higher than that of other scales, suggesting a general lack of self-observation and severe aversion to self-observation in students. This evidence could imply a general tendency towards closure in our studied student sample during the lockdown period. Did the quarantine situation increase the *Defensive attitude* of students during online education? Moreover, the correlation analysis results showed no significant values between QBS scales and RCMAS-2 scales in the Total (45) Junior student group. However, considering HL and NH separately, results showed, in the HL group, a negative correlation between *Defensive Attitude (RCMAS)* and RWT (QBS) ($r = -0.35$, $p < 0.05$) not present in the NH group. Whereas, the latter group revealed a strong positive correlation between *Defensive Attitude* and RWC ($r = 0.85$, $p < 0.05$) and between *Defensive Attitude* and SE ($r = 0.72$, $p < 0.05$). It is well known that the teacher-student relationship is predictive of classroom wellbeing (Murray & Pianta, 2007; Spilt et al., 2011; Wolters et al., 2012). Moreover, the relationship with the teacher is potentially even more significant for the wellbeing of students with disabilities (Murray & Greenberg, 2001; Murray & Pianta, 2007). For deaf children in special education schools, a more positive relationship with the teacher increases wellbeing in school in both Grades 6 and 7 (Wolters et al., 2012). We can hypothesize that the negative correlation between *Defensive attitude* and RWT may represent a synergistic element in the wellbeing of the HL child during the quarantine school isolation period. Regarding the correlations in the NH group between *Defensive attitude* and RWC and SE, respectively, our results appear to be in line with the literature that has shown how children and adolescents rely heavily on the evaluation of others for self-assessment. From these assertions, regarding our results, one could possibly advance the hypothesis that during the pandemic, the insecurity due to the lack of physical and nonvirtual regularity of the relationship with peers has increased in the NH population a defensive attitude in an attempt to preserve the quality of their relationship with peers and the maintenance of self-esteem. Despite previous evidence (P. K. Bender et al., 2012), in the present study sample, Gender did not significantly influence clinical anxiety features (Table 1 [**B**]). This lack of difference could be because anxiety was assessed via self- reporting measures (online survey). In fact, although this methodology is common in the literature (e.g., Garnefski et al., 2005; Martin & Dahlen, 2005), self-reporting measures may cause some bias in the way they require respondents to report on their behavior. It is worth noting that, on average, girls obtained higher scores in total anxiety than males (see Table 1 [**B**]) as reported in the normative groups (Reynolds et al., 2012).

Furthermore, the higher value of *Defensive attitude* compared to the other RCMAS-2 scales, supports the idea that the student groups (regardless of Gender and Hearing factors) concealed some anxiety symptoms, at least during lockdown. Will the strong defensive attitude persist even after the restoration of face-to-face teaching? Did quarantine increase defensive attitudes in NH or HL children more than with face-to-face teaching? Once again, we may possibly answer these intriguing questions in a future study involving regular teaching conditions.

4.4 | Parents

With regard to the assessment of the overall parent population, low symptoms of Trait anxiety and mild symptoms of State anxiety are observed in respect of the Italian normative standard (Spielberger, 2018). It is possible to suggest that, in line with recent studies (Marchetti et al., 2020; Mazza et al., 2020; Prete et al., 2020), the general population's level of anxiety has risen due to Covid-19 fear and uncertainty.

We observed a significant global difference between Trait and State anxiety scores in parents: the latter are higher (Figure 6). According to Spielberger (1966; 1979), State anxiety reflects the transitory emotional state of human reactions directly related to adverse situations in a specific moment of the life. In contrast, Trait anxiety refers to a trait of personality, describing individual differences related to the predisposition to respond anxiously to certain situations. Our results show ho1w the adverse conditions faced by these parents during lockdown due to Covid-19, significantly influenced their levels of anxiety. Moreover, this evidence is in line with recent studies that reported a high level of anxiety among Italian adults during the pandemic

(Casagrande et al., 2020; Cellini et al., 2020; Rossi et al., 2020), supporting the hypothesis that during quarantine, Italian parents evaluated the pandemic as severe, showing a realistic perception of the critical situation. In keeping with our research proposal, we conducted additional analysis to understand whether other factors, in addition to the quarantine period, influenced the anxious parental state.

No differences emerged between parents with a HL or NH child. Studies posit that the levels of parental anxiety affect the physical and mental development of children (e.g., Kennedy et al., 2009; Ramchandani et al., 2005; Sprang & Silman, 2013). It is therefore sadly imaginable how parents of SN children (and so HL children) who have to undertake the tasks of childcare, training, rehabilitation and learning during the pandemic would pose an immense challenge for them.

Exploring parental anxiety under stress and the corresponding influencing factors prevalent during Covid-19 will help healthcare professionals to provide targeted guidance and assistance. From this perspective, it is reasonable to believe that parents of HL children will have been more stressed than usual during the pandemic because they would have had to take care of their children's education and overall wellbeing, which eventually leads to an increase in anxiety state levels.

Our results are partially in line with a fascinating recent study (Ren et al., 2020) about the state anxiety of parents of SN children during Covid-19 that shows significant differences in the results amongst parents. Authors found that the state anxiety score was significantly higher in parents of children with autism than in parents with visual impairment, while they have not observed any differences amongst parents of autistic, intellectual, and hearing-impaired children. However, the study design of Ren and colleagues (2020), did not include a group of parents of healthy children, and so no comparison with a control group has been assessed. Therefore, in an evaluative comparison between our results and those of Ren (2020) it is not possible to test whether additional factors could contribute to any lack of significant differences between HL parents and NH ones: for example, in the identification of protective factors for state anxiety in parents with SN children or the presence of risk factors in parents with healthy children. With respect to the question of whether there is the existence or absence of additional factors that could justify the lack of significant differences between a parent of a HL student and those of NH students, we have further deepened the analysis with additional variables (Gender, Income, Education).

The Gender of the child does not affect the level of parental anxiety. Some studies on younger children found that same-gender parent-child dyads demonstrate a strong relationship between parent and child psychopathology (e.g., Ensminger et al., 2003; Wahl & Metzner, 2012), whereas studies on emerging adults have indicated that opposite gender parent-child dyads tend to have the strongest associations between parent and child psychopathology (McKinney & Brown, & Malkin, 2018; McKinney & Kwan, 2018; Walker & McKinney, 2015). However, although the literature emphasizes the correlation between mental disorder within parent-child dyads, few studies have delved into the differences in parental state anxiety based on child gender. As noted for the Auditory Condition factor, we did not find a significant effect due to the Gender of the child on parental State and Trait anxiety. However, we observed significant correlations between child anxiety and parent anxiety in both NH and HL populations (Table 4). It is interesting to note how the Total anxiety in students is correlated with both State ($r = 0.37$, $p < 0.05$) and Trait anxiety of parents ($r = 0.35$, $p < 0.05$) in the HL group and not in the NH group. Moreover, for all RCMAS-2 scales, positive correlations with State and Trait anxiety are observed both in NH and HL groups but with higher values in NH than in the HL group. Thus, although State and Trait anxiety may be concomitant factors with the child's Total anxiety, the components of this psychic condition (in particular *Defensive attitude and Concern*) seem to be most strongly correlated in the NH group ($0.45 \geq rNH \leq 0.67$, $0.35 \geq rHL \leq 0.41$; $p < 0.05$). We can speculate that the tendency to conceal certain aspects of oneself and appear differently from how one is, would seem to be more aligned within the NH population to parental anxiety status during quarantine for Covid-19.

Studies investigating the risk factors for anxiety caused by the Covid-19 outbreak reported that anxiety or depression were associated with loss of income due to the pandemic (Hyland et al., 2020). Another study found no significant association between occupation, income, and anxiety during this challenging period (Blbas et al., 2020).

Moreover, as observed in a recent review by Brooks et al. (2020), financial loss can be a severe problem during the pandemic. Authors reported that economic loss due to quarantine created serious socioeconomic distress (Pellecchia et al., 2015) and was found to be a risk factor for symptoms of psychological disorders (Mihashi et al., 2009) and resulted in both anger and anxiety several months after quarantine (Jeong et al., 2016) Recently, L. Chen et al. (2021) showed a trend of negative correlation between income levels and STAI-Y scores, finding also that the incidence of severe anxiety and STAI-Y scores in low-income groups significantly increased during the quarantine period. However, to date, few studies (if any) have focussed on the factors that may affect the anxiety of quarantined parents with HL children. Although pandemic diseases, as seen previously, were found to have been associated with high levels of anxiety as recorded in recent literature, the mechanism underlying specific processes is still unclear, especially, in disability affected populations. For example, Ren et al. (2020) found that parents of SN children with a monthly family income above 15k dollars have the lowest levels of anxiety. Our results do not show any differences in parent State and Trait anxiety with different family Income levels and this evidence is also confirmed by the absence of correlation between anxiety and family income in HL and NH parent groups (Table 3 [**B**]).

We can speculate that in the participant groups the lack of differences in anxiety (S and T) based on Income level is due to the fact that risk and protective factors modulating anxiety are different, based on whether you have a NH or HL child. These results strongly support the idea that a re-evaluation of the impact of Income on anxious symptomatology is necessary. In fact, since 2020, Covid-19 has crucially affected the development of economies and wider society, in Italy as well as throughout the world (ISTAT, 2021). Many families have lost their jobs and have had reduced standards of living (MEF, 2021): it is clearly recognized that lower levels of household income are associated with several mental disorders (Sareen et al., 2011). Therefore, an assessment of the anxious state of parents is essential for targeted interventions especially in families with impaired children.

Concerning the Education Level of parents, Ren and colleagues (2020) highlighted that during the Covid-19 pandemic, parents of SN children with a College education or above experienced a lower level of state anxiety than those who only reached Senior High School. Similarly, mothers of disabled children with lower educational levels had the most elevated Trait anxiety (Bumin et al., 2008). Moreover, a study conducted in Australia during the equine flu epidemic, found that minor educated groups were at greater risk of mental distress (Taylor et al., 2008). In contrast to these studies, but in line with Mappa et al. (2020), who observed that a higher educational status was associated with increased prevalence of anxiety, our results do not show significant differences regarding parent's education level. Although level of education does not seem to affect the anxiety level in adult participants, the only group that showed a significantly strong negative correlation between State Anxiety and Education Level were the parents of NH children ($r = -0.66$, $p < 0.05$). We can hypothesize that education was a protective factor against anxiety in the parent-child dyad during the quarantine period because parents with higher educational qualifications are more likely to learn and master the skills necessary to cope with their anxiety, avoid experiencing its adverse effects and passing them on to their children. Moreover, parents of HL children, showed significative positive correlation between State anxiety and level of *Concern* about the school closures (Table 3 [**B**]).

5 | CONCLUSION

In conclusion, the intent of the COCLOVID study was to open a small window on our collective understanding of the educational and psychological wellness of children experienced during the complex pandemic period currently being faced and which may be particularly difficult for students with hearing difficulties and their families.

The results of the present study allow us to answer initial questions as follows:

(1) In our sample participating in online education during the Covid-19 pandemic and having hearing difficulties or being a parent of a child with hearing difficulties did not seem to affect School Wellbeing.

(2) Although much of the literature generally reported significant psychological differences between students with SNs and their peers, results of the present study do not show macro differences between hearing impaired and normal hearing students for anxiety levels experienced during the lockdown. At the same time, this lack of differentiation based on deafness was also present amongst parents who nevertheless also showed moderate anxiety symptoms. It is possible to suggest that the level of anxiety of parents may have risen due to Covid-19 and not to their children's impairment.

(3) Normal hearing parent-child dyad seems to show the strongest correlation in terms of parental anxiety and children's defensive attitude. Different psychological costs between children, with or without hearing impairments, can be observed in term of the relationship between defensive attitude and relationships with classmates and teachers.

6 | LIMITATIONS

We are aware that the use of an online tool is not the optimum methodological choice available especially when the objective is the assessment of sensitive variables such as psychological ones. However, this choice was necessary to reach participants in a short period of time and during a pandemic, when face-to-face contacts were forbidden or severely restricted. Furthermore, although bias can affect any survey (Pierce et al., 2020), the methodology adopted in our study made it possible to avoid interpretative bias due to participants' hearing difficulties. Additionally, although the results are limited by the size of the sample observed they appear to be a relevant contribution to the debate on the impact of online education, as faced by students around the world. A final limitation of the study, shared with most existing empirical studies on Covid-19, is the difficulty of parsing causal relationships due to collecting self-reporting measures with no prepandemic baseline available. A future comparison with the results of an investigation undertaken in a normal educational situation with in class learning may provide support for a causal analysis and could give direction for a targeted intervention on the wellbeing of students and their families in the broader context on an effective inclusive school.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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Stefania Di Biasi, Diana Di Pietro

Bambini e adolescenti "a distanza"

Il disagio psichico e l'emergenza psicopatologica durante la Pandemia da Covid-19

alpes

Psiche e dintorni

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SOMMARIO

COME (SI) SENTONO I RAGAZZI?
Riflessioni su Didattica a Distanza e Sordità ai tempi della pandemia

Bianca M.S. Inguscio, Maria Nicastri, Ilaria Giallini, Antonio Greco, Fabio Babiloni, Patrizia Mancini, Giulia Cartocci

> "Presta a tutti il tuo orecchio, a pochi la tua voce"
> (W. Shakespeare)

Un colpo d'ala irruento, sebbene a distanza

Alle porte della Primavera 2020 l'Oms (Organizzazione Mondiale della Sanità) ha dichiarato il Sars-Cov-2 essere un virus pandemico (πανδή μιος *-pan demos*: di tutto il popolo[1]). Un'ala di ombra che, indifferentemente dalle singolarità individuali, con un battere irruento e cupo ha sovrastato l'umanità portando, tra continenti, stati, regioni città, famiglie, persone, un'imposta gabbia di omogeneità nel sentire il mondo esterno. Difatti, per contenere la trasmissione del virus, il distanziamento sociale è stato introdotto a livello mondiale e di conseguenza, i portoni delle scuole, spazi elettivi non solo per l'istruzione ma, soprattutto, luoghi di formazione alla vita, rapidamente sono stati sbarrati. La preoccupante situazione sanitaria ha fatto sì che oltre il 90% dei bambini e dei ragazzi iscritti a scuola in tutto il mondo abbia dovuto abbandonare il proprio banco (United Nations Educational, Scientific and Cultural Organization, Unesco 2020), con la consapevolezza, emersa anche dall' affermazione del Direttore Generale Audrey Azoulay di come "… *Mentre le chiusure temporanee delle scuole come risultato di crisi sanitarie e di altro tipo non sono purtroppo nuove, la scala globale e la velocità dell'attuale perturbazione educativa non ha eguali e, se prolungata, potrebbe minacciare il diritto all'istruzione*[2]*….".* (Mia traduzione*).*

L'Italia è stato il primo paese in Europa ad attuare il *lockdown* (confinamento, NdA.) a livello nazionale durante il quale bambini, ragazzi e le loro famiglie hanno vissuto in totale isolamento per circa due mesi fino al 3 maggio 2020, mentre le scuole sono rimaste chiuse fino a settembre.

Al fine di mitigare gli effetti della brusca interruzione della didattica i paesi hanno rapidamente deciso di avviare modalità di apprendimento a distanza erogate attraverso diversi canali, da piattaforme online a programmi televisivi, radiofonici e materiali cartacei[3]. In Italia, il Ministero dell'Istruzione dell'Università e della Ricerca (Miur, 2020) ha istituito l'obbligo di attivare la Didattica a Distanza (DaD), intesa come una modalità di didattica che permette a studenti ed insegnanti di proseguire il percorso di formazione e apprendimento anche se "fisicamente" distanti.

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[1] Enciclopedia Treccani Online, consultabile al seguente link https://www.treccani.it/vocabolario/ricerca/pandemia/

[2] Testo originale: "…*While temporary school closures as a result of health and other crises are not new unfortunately, the global scale and speed of the current educational disruption is unparalleled and, if prolonged, could threaten the right to education….".*

[3] Per un approfondimento sull'impatto globale della DaD si veda l'interessante report di Conto et al. 2021 per l'Unicef.

A supporto concreto del modello di DaD per tutti i discenti italiani, sono stati stanziati nell'anno ampi finanziamenti per fornire dispositivi digitali e di connettività a tutti i bambini e ragazzi provenienti da contesti economici svantaggiati e supporti all'attività ed alla formazione digitale degli insegnanti; permettendo di raggiungere così, nel nostro paese, potenzialmente 6.7 milioni di studenti su 8.3[4] malgrado l'Istituto Nazionale di Statistica (Istat, 2020) abbia stimato che circa 3 milioni di studenti siano stati esclusi dalla didattica *online* per la carenza di connessione o di adeguati strumenti. Tuttavia, sebbene vi sia una pletora di tecnologie in continuo ed interrotto sviluppo per permettere un accesso scrupoloso e costante alla formazione, spesso sono le stesse tecnologie a creare difficoltà per gli studenti.

Bastano uno schermo ed una rete di connessione digitale per ricreare l'ambiente costruttivamente disarmonico di una classe? La *rete* collega punti separati, intrappola pesci pescati, o riesce a coinvolgere empaticamente chi vi prende parte? Lo s*chermo*, finestra sul mondo di questo anno passato tra le mura domestiche, permette una effettiva percezione del soffio del vento, del sole che brilla e della cupa tempesta garantendo una relazione costruttiva con ciò che osserviamo davanti a noi o semplicemente, *scherma* noi stessi da una realtà sensoriale filtrata e subdola? Comenio, studioso del Rinascimento, sosteneva che il fondamento dei processi di apprendimento risiede nel "… presentare direttamente le cose sensibili ai sensi, sicché non possano non essere comprese… niente è nell'intelletto, che prima non sia nel senso…" (Comenio, 1969, p 563).

Forse, il più grave errore che si possa commettere nell'avvicinarsi ad avanzare considerazioni sulla DaD, è davvero quello di pensare che in fondo, sia identica a quella svolta nelle aule solo con una *webcam* ed uno schermo in mezzo, come osservano Bruschi e Perissinotto (2021). Inoltre, chiusi nelle loro stanze, gli studenti vivono un mondo mai vissuto senza precedenti, dove lo spazio condiviso con l'Altro, delimitato da uno schermo bidimensionale, blocca l'interazione, la partecipazione diretta cancellando o diminuendo sostanzialmente i feedback costruttivi che permettono una crescita.

È chiaro come la chiusura delle scuole abbia stravolto come mai sino ad oggi le vite degli studenti e delle loro famiglie imponendo ai ragazzi una modalità di apprendimento totalmente nuova ed ai genitori un ruolo di supporto da pseudo-educatori. Con l'aumento dell'uso di internet durante il *lockdown*, i genitori hanno giocato (e giocano tutt'ora) un ruolo ancora più centrale sulla scacchiera educativa. È quindi comprensibile che i genitori possano condividere le preoccupazioni dei loro figli riguardo alla DaD o sentirsi sopraffatti da questo nuovo compito Il progetto KiDiCoTi coordinato dal Centro Comune di Ricerca della Commissione Europea ha evidenziato come, mentre più di un bambino o ragazzo su tre ha riferito di preoccuparsi di non essere in grado di stare al passo con il lavoro scolastico a causa dell'isolamento, più della metà dei genitori era 'preoccupata' o 'molto preoccupata' dell'impatto negativo

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[4] https://scuola24.ilsole24ore.com/art/scuola/2020-03-26/didattica-digitale-raggiunti-67-milioni-studenti-sugli-83-milioni-complessivi-164052.php?uuid=ADex49F&refresh_ce=1

che la pandemia avrebbe potuto avere sull'istruzione dei propri figli. Risulta inoltre incoraggiante osservare che il 92% dei genitori ha dichiarato di possedere sufficienti competenze digitali per sostenere i propri figli nella DaD, tuttavia il 28% dei genitori ha anche affermato di non aver avuto abbastanza tempo per sostenere i propri figli nella didattica durante l'isolamento ed il 27% di non essersi impegnato in attività educative extra con i propri figli al di fuori di quelle erogate dalla scuola (Mascheroni et al. 2021).

È facilmente immaginabile come l'isolamento, la mancanza di supporti esterni all'organizzazione delle *routine* familiari, abbiano avuto ripercussioni sia sullo sviluppo sociale, psicologico ed educativo di bambini ed adolescenti (come ampiamente approfondito nei capitoli precedenti) sia sul reddito e la produttività dei genitori che hanno spesso dovuto ridurre l'attività lavorativa per prendersi cura dei figli. Così, il sostanziale e repentino cambiamento dell'ambiente di vita, delle abitudini giornaliere e dei contatti sociali, fondamentali per promuovere il benessere psicofisico e la resilienza ad eventi traumatici, ha travolto milioni di studenti, costretti a sospendere, oltre all'attività didattica, anche le attività sportive e culturali[5].

Integrare le diversità per uno sviluppo comune

L' Unesco nella recente Giornata Internazionale dell'Educazione[6] ha ribadito come l'educazione sia un diritto umano, un bene ed una responsabilità pubblica oltre a sottolineare come la situazione pandemica abbia reso l'umanità ancor più consapevole di quanto la scuola sia più di un semplice luogo di apprendimento, ma un luogo che offre protezione, benessere, cibo e libertà.

Considerando la *ritualità* un aspetto fondante che caratterizza l'individualità e lo sviluppo di ogni essere umano (Erikson 1950; 1966) e di come la *routine scolastica* sia un pilastro fondamentale per il sano sviluppo del fanciullo, è tristemente immaginabile come la situazione di chiusura e l'imposizione della DaD abbiano avuto un impatto amplificato sui ragazzi con bisogni educativi speciali (BES)[7], in particolare con disabilità sensoriali, come la sordità. Precedenti ricerche mostrano difatti come le difficoltà uditive si associno ad un'eterogeneità di esiti nello sviluppo cognitivo, sociale ed emozionale (Kral e Stato, 2020). Inoltre, per il bambino sordo, la scuola "…rappresenta una fondamentale occasione di apprendimento e di condivisione dell'esperienza con coetanei ed adulti diversi dai familiari…" (Bosco, 2013, pag. 99).

Di conseguenza, la disabilità uditiva non li pone di per sé a maggiore rischio di contaminazione in questa tempesta pandemica ma li espone al rischio di ulteriori violente correnti mosse dalle discriminazioni e

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[5] Per un approfondimento dell'impatto della pandemia sui settori culturali e ricreativi si rimanda a Oecd, 2020.

[6] https://en.unesco.org/commemorations/educationday

[7] Il concetto di Bisogno Educativo Speciale (BES)/Special Education Need (SEN) appare nei documenti ufficiali Unesco nel 1997, nella legislazione del Regno Unito nel 2001 (Special Needs and Disability Act) e nei documenti dell'Agenzia Europea per lo Sviluppo dell'educazione per i bisogni speciali nel 2003, come tendenza a considerare soggetti con BES anche altre persone in età evolutiva che manifestino difficoltà di apprendimento e di comportamento diverse dalla disabilità (Ianes e Cramerotti, 2013, p. 19).

dalle barriere all'informazione, alla salute ed all'inclusione sociale e scolastica. A causa delle limitazioni sensoriali, gli individui sordi, in particolare i bambini, possono sviluppare dei tratti psicologici che li rendono più vulnerabili rispetto ai normoudenti (Al Majali e Alghazo, 2021) e diversamente resilienti alle situazioni avverse. Inoltre si osserva che la prevalenza degli studi pubblicati sugli effetti della pandemia sono focalizzati sullo status psicologico di soggetti senza problematiche uditive e rare sono le ricerche che affrontano le difficoltà riscontrate dagli studenti sordi nell'era covid e dei genitori di figli con bisogni speciali. Nel panorama italiano, il *case study* di Tomasuolo e colleghi (2021), ha esplorato nel complesso l'impatto della crisi pandemica sulla comunità sorda italiana intesa come minorità linguistica (segnante), mentre, a nostra conoscenza, attualmente si nota un vuoto di contributi scientifici italiani focalizzati sul vissuto psicologico degli alunni e studenti sordi e le loro famiglie durante la didattica a distanza.

Pertanto, alla luce di quanto esposto, è intuibile come, in silenzio, molti bambini affetti da ipoacusia[8] ed i loro genitori abbiano affrontato la complessa e poliedrica esperienza della didattica a distanza, riscontrando sia difficoltà che benefiche, nuove, scoperte.

Durante le lezioni *online*, molteplici problemi tecnici possono insorgere a causa dei dispositivi uditivi, come gli impianti cocleari e le protesi acustiche[9], che rispettivamente permettono e coadiuvano l'ascolto nello studente sordo ed ipoacusico; oppure della piattaforma digitale utilizzata la quale non sempre permette di cogliere al meglio i movimenti delle labbra, essenziali a chi, come l'ipoudente, spesso si affida alla lettura labiale per seguire il filo comunicativo. La lettura delle labbra, difatti, è pienamente efficiente solo quando le condizioni sono ideali. Ci sono spesso ostacoli come i baffi, la scarsa illuminazione, il luogo e la posizione sbagliata del parlante, la celerità nell'espressione parlata, gli accenti stranieri, gli omofoni linguistici, che possono inficiare il messaggio comunicativo. Tuttavia, per alcuni alunni ipoudenti le lezioni svolte davanti ad uno schermo sembra siano state più efficaci a livello didattico, come si può evincere dalle reali parole di alcuni ragazzi di seguito riportate[10].

"… *Con la didattica a distanza uso delle specie di cuffie che mi permettono di sentire bene le voci di tutti, il problema è che quando i miei compagni parlano insieme, non riesco a capire nulla. Preferirei le lezioni in presenza per poter interagire con i professori e con gli amici in classe, ma l'uso delle mascherine è una barriera che mi impedisce di vedere il labiale ed attutisce il tono della voce quindi, alla fine, per me è meglio la DaD".* Maria, quarta superiore.

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[8] In audiologia i deficit della funzione uditiva vengono definiti ipoacusie. A seconda del livello di perdita della percezione uditiva (misurata in dB) si identificano diversi livelli di ipoacusia (da lieve = perdita della percezione dei suoni tra 25-34 dB a profonda=perdita della percezione dei suoni da 95 dB). Per un approfondimento si rimanda a Cianfrone, 1986.

[9] Per un approfondimento dei devices tecnologici utilizzati a supporto delle ipoacusie si rimanda a Ambrosetti et al. 2018.

[10] Testimonianza raccolte dall'associazione fiorentina "Io parlo" pubblicate al seguente link https://www.redattoresociale.it/article/notiziario/_noi_alunni_sordi_non_leggiamo_il_labiale_dei_maestri_con_la_mascherina_

"…*A me piace molto andare a scuola e mi piacerebbe tornare in aula con i miei compagni di classe ma se i professori portano la mascherina non riesco a comprendere quasi niente. Da casa, con le cuffie e gli apparecchi riesco a guadagnare ma mi manca tantissimo poter interagire con i miei compagni*". Agnese, prima superiore.

"… *A me non piace la didattica a distanza, faccio fatica a concentrarmi con le lezioni al computer. A scuola i prof solitamente non usavano la mascherina. Sulla cattedra era stata messa una barriera-schermo in plexiglas e così loro stavano lì dietro ed io potevo seguire le lezioni senza molte complicazioni, Il plexiglas un po' attutisce il tono della voce ma vedo bene il labiale è un grande aiuto". Giovanni, seconda media.

Da questi frammenti espressi, si scorge il quadro di complessità all'interno del quale gli studenti con problematiche uditive si sono mossi e devono tuttora muoversi per vivere un benessere scolastico, essenziale per il loro sviluppo e la loro crescita. Poche parole, come silenziose pennellate, suggerenti come, aldilà dello schermo, si schiudano esistenze complesse e difficoltà multisfaccettate, purtroppo non affrontabili e risolvibili unicamente con l'azione del legislatore.

I vissuti del singolo sono sfumature infinite: allegrie, tristezze, angosce, gioie espresse che, da schermi sebbene ad alta risoluzione e forte connessione (nei migliori dei casi), non possono essere colte. Una tavolozza di tempere molteplici, una *tela* impenetrabile ai cromatismi della vita.

Lo *schermo* permette l'accesso ad un canale comunicativo ma, come osserva Ammaniti "…Gli scambi attraverso lo schermo inevitabilmente provocano distorsioni e ritardi nella percezione proprio perché la stessa codifica digitale comporta alterazioni, adattamenti e sintetizzazioni complesse che creano artificiosità relazionali. Congelamenti delle immagini, nebulosità, blocchi e scatti, ridotta sincronia fra video e audio sono fenomeni più che frequenti che generano confusioni nella percezione e negli stessi codici sociali…" (Ammaniti, 2020a).

Come può aver vissuto questa esperienza uno studente ipoudente che di per sé abita un mondo percettivo non istantaneamente sincronizzato con il mondo esterno, dove un suono arriva con ritardo e con ritardo l'emozione può essere decodificata e compresa? Quanto lo schermo può essere considerato uno specchio trasparente che permette una comunicazione empatica? È realmente capace di favorire una didattica inclusiva che, oltre la *distanza*, renda la lezione *vicina* agli studenti ed ai loro profondi bisogni?

Come osserva Stella "… quello che manca ai bambini è certamente il contatto e il lavoro di gruppo: la ridondanza dei contenuti proposti dal docente, e realizzata anche con l'intervento di tutti i bambini, ha un effetto di rinforzo dell'apprendimento che non si riesce ad ottenere quando ciascuno è davanti al suo computer…" (Stella, 2021, pag.55).

Non vi sono risposte chiare alle infinite domande sulla DaD che stanno emergendo nel corso della pandemia. Non vi sono evidenze a riguardo dei vissuti psicologici delle famiglie con studenti con difficoltà uditive. La stessa istruzione, che nel nostro Stato riveste un ruolo cogente, con l'avvento dell'estranea modalità didattica *online* è apparsa come una nuova città, davanti alle porte della quale anche il più esperto viaggiatore si è sentito intimorito…senza esperienza. Ogni studente, insegnante, genitore nel corso della didattica durante il *lockdown* avrà percepito internamente, ricordando Italo Calvino e le sue Città Invisibili "…l'estraneità di ciò che non sei più o non possiedi più che t'aspetta al varco dei luoghi estranei e non posseduti…".

Inclusione o esclusione

È bene sottolineare in questa sede come la didattica inclusiva sia uno degli obiettivi trainanti il sistema scolastico italiano. Lo stesso Ministero dell'Istruzione dell'Università e della Ricerca sottolinea che *"…l'integrazione scolastica degli alunni con disabilità costituisce di fatto un punto di forza della scuola italiana, che vuole essere una comunità accogliente nella quale tutti gli alunni, a prescindere dalle loro diversità funzionali, possano realizzare esperienze di crescita individuale e sociale…*"[11]. L'inclusione scolastica ha come obiettivo l'integrazione di tutte le necessità degli studenti con disabilità e/o bisogni educativi speciali con l'obiettivo di permettere loro lo sviluppo dell'apprendimento e delle condizioni necessarie al loro sviluppo sociale. Indicatori efficaci di una didattica inclusiva possono essere considerate variabili psicologiche quali attitudini, preoccupazioni, senso di autoefficacia[12].

Sebbene la didattica *online* rappresenti un nuovo orizzonte per lo sviluppo e l'integrazione in soggetti con ipoacusia severa e profonda nel panorama internazionale, esigui sono attualmente gli studi che indagano l'impatto e l'efficacia della DaD in questo tipo di popolazioni[13]. Pertanto, ricollocando il discorso lungo il sottile sentiero del presente contributo, è evidente come quando la valutazione dell'efficacia di un modello di didattica inclusiva (a distanza) è discusso, la dimensione psicologica sia essenziale specie in un paesaggio emergenziale complesso come quello che la nostra comunità, in particolare famiglie con figli con disabilità sensoriali, sta attraversando.

Silenziose sinergie uditive

Un piccolo contributo al ridimensionamento del vuoto italiano di evidenze relative al benessere psicologico percepito durante la DaD dagli studenti ipoudenti, è stato quello realizzato nell'ambito del progetto COCLOVID (Valutazione della Didattica Online nel corso della quarantena per COVId in

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[11] https://www.miur.gov.it/alunni-con-disabilita.

[12] Sul costrutto di autoefficacia si rimanda a Bandura, 2000.

[13] Per un approfondimento si veda ad esempio Hashim et al. 2013.

bambini portatori di impianti COCLeari e genitori), promosso dal Centro Impianti Cocleari del Policlinico Umberto I in sinergia con i Dipartimenti di Organi di Senso e di Medicina Molecolare della Sapienza, Università di Roma (Inguscio et al. 2020). In questo ambito, lo sguardo di approfondimento dei ricercatori è stato rivolto all'indagare come bambini ed adolescenti con dispositivi acustici si siano adeguati alla DaD durante l'isolamento e quali siano stati i loro vissuti interpersonali con l'obiettivo di svelare criticità e/o potenzialità di essa. Attraverso un questionario diffuso online, comprendente valutazioni qualitative e quantitative, è stato possibile cogliere indirettamente i vissuti degli studenti e dei loro genitori, illuminando, sebbene con limitato spiraglio, la vita oltre lo schermo.

Il questionario è stato compilato su base volontaria da più di sessanta coppie di genitori e figli, di cui circa la metà affetti da deficit uditivi. Oltre a richiedere informazioni sociodemografiche ed opinioni sulla DaD, il questionario ha permesso di indagare, attraverso strumenti psicometrici validati internazionalmente, aspetti psicologici quali il benessere scolastico dello studente percepito direttamente dal figlio e secondo la prospettiva genitoriale e la sintomatologia ansiosa dei partecipanti. I risultati salienti[14] hanno fatto emergere alcuni nuclei di riflessione.

La maggior parte dei genitori, indipendentemente dall'avere un figlio ipoacusico, ha riportato di sentirsi 'abbastanza' preoccupato per la chiusura delle scuole ritenendo la DaD 'abbastanza' utile. Differentemente, un numero maggiore di genitori con figli ipoudenti ha affermato di essere 'estremamente preoccupato' per la chiusura delle scuole, ritenendo la DaD 'estremamente utile'. Per quanto riguarda la prospettiva dei discenti, la maggior parte di loro ha considerato le lezioni *online* 'abbastanza piacevoli', solo alcuni studenti sordi le hanno considerate 'estremamente piacevoli'.

Nel complesso la didattica a distanza è stata considerata 'normale' dalla maggior parte degli studenti, 'stancante' da un numero superiore di ragazzi sordi. Per questi ultimi, l'elemento segnalato che non permetteva loro di seguire bene le lezioni è stato la 'velocità nel parlato dell'insegnante'.

Già da queste generiche osservazioni qualitative emerge come la sordità comporti un differente giudizio nei confronti della didattica *online* sovrastante differenti vissuti. La somministrazione dei questionari psicologici ha permesso una valutazione quantitativa del *benessere scolastico* e dell'*ansia*.

Globalmente, contrariamente a quanto ipotizzabile, i dati hanno mostrato come il deficit sensoriale non abbia un impatto significativo sul benessere scolastico e sulla sintomatologia ansiosa degli studenti né dei loro genitori. Tuttavia, in merito all'ansia, emerge uno spiccato *atteggiamento difensivo*[15] in tutti i ragazzi intervistati, segnale di una tendenza a celare alcuni aspetti di sé cercando di mostrarsi in modo migliore agli occhi degli altri. Questo atteggiamento di auto-protezione, immaginabile in un

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[14] Per una descrizione completa dello studio COCLOVID si rimanda ad Inguscio et al. 2021 - *submitted*)

[15] Per atteggiamento difensivo si fa riferimento alla scala del questionario RCMAS-2, strumento self-report per la valutazione dell'ansia in età evolutiva, che permette di identificare se il soggetto è disposto ad ammettere le imperfezioni quotidiane come parte dell'esperienza comune (Reynolds et al. 2012).

contesto in cui la relazione con l'altro è filtrata (o meglio …*schermata*) dai sussidi tecnologici di connessione con la classe potrebbe, se prolungato nel tempo, inficiare la costruzione ed il mantenimento della relazione con i pari. D'altro canto, è interessante notare come mentre negli studenti normoudenti vi sia una infelice relazione positiva tra l'atteggiamento difensivo e la relazione con i compagni di classe, negli ipoudenti è la relazione con gli insegnanti ad essere legata (in questo caso negativamente) all'atteggiamento difensivo. Queste evidenze potrebbero condurre ad ipotizzare che nel periodo di DaD in isolamento per la Covid-19, il rapporto con l'insegnante sia stato un fattore costruttivo e protettivo nei confronti di manifestazioni ansiose per i ragazzi ipoudenti, fattore non emerso tra gli studenti normoudenti dove invece si osserva un incremento di atteggiamento difensivo legato alla relazione con i compagni. È importante tenere in mente questa criticità che potrebbe essere un ulteriore segnale di perdita di fiducia gli uni negli altri tra compagni di classe (elemento che emerge anche nel documento dell'Unicef "Protezione dei bambini durante la Pandemia di coronavirus (2020).

Il campione dei genitori analizzato ha mostrato un medio livello di ansia dovuto alla situazione di isolamento correlato ad alcune variabili concernenti l'ansia del figlio. Questi risultati offrono un piccolo, importante, spaccato della situazione delle famiglie, in linea con l'ampia indagine svolta dall'Istituto Gaslini in collaborazione con l'Università di Genova sullo stato psicologico di bambini e famiglie (Uccella et al. 2021) che ha mostrato come la situazione di confinamento abbia avuto ripercussioni significative sulla salute emozionale e psichica dei genitori e dei bambini. Lo stato ansioso che ha accomunato le diadi partecipanti allo studio COCLOVID riporta a quei "…grumi emotivi degli adulti che entrano nel mondo mentale dei bambini creando tensioni e risonanze che si ripercuotono sul sistema dei neuromodulatori cerebrali, sul sistema neurovegetativo e sullo stato psicologico…" (Ammaniti, 2020b, p. 49).

Considerando la disabilità sensoriale nel campione dei genitori, è interessante osservare che sebbene non emergano differenze significative nel vissuto ansioso del genitore con figlio sordo o normoudente, come invece sembrerebbe suggerire la letteratura (in particolare lo studio di Ren e colleghi 2020 sullo stato ansioso di genitori con bambini con bisogni speciali), solo l'ansia del genitore con figlio ipoudente si correla maggiormente con la sua preoccupazione per la chiusura delle scuole. Questa osservazione è tristemente accettabile considerando come un genitore di un bambino ipoudente nel periodo di confinamento abbia dovuto farsi carico oltre della cura consueta del figlio, di tutti quegli aspetti psicoeducativi e motori che, in condizioni normali, vengono generalmente delegati ad una rete di strutture di supporto esterne. Asbury e colleghi (2020) d'altronde hanno rilevato un profondo sentimento di perdita delle *routine*, della rete e delle strutture di supporto e dei servizi specialistici, tra le categorie più ricorrenti nelle testimonianze dei genitori con bambini con bisogni educativi speciali, senza focalizzare tuttavia sulla sordità.

I risultati preliminari dello studio COCLOVID, sebbene da un lato non identifichino un netto confine dell' impatto della DaD sul benessere psicologico e scolastico tra studenti normo ed ipoudenti e la completa inclusione di quest'ultimi nell'attuale paradigma educativo, suggeriscono la necessità di una valutazione *follow -up* (ad oggi avviata dal gruppo di ricerca del Centro Impianti Cocleari del Policlinico Umberto I di Roma) con la riapertura delle scuole ed il conseguente ritorno ad una didattica in presenza.

Concludendo, il presente contributo non pretende di offrire una meccanicistica risposta e soluzioni alle infinite eco di domande e riflessioni scaturenti dallo spaccato dell'impatto della DaD su studenti normo ed ipoudenti; ma vorrebbe non rendere sorda la voce di questa eco che, se ascoltata, potrebbe divenire fiaccola illuminante silenziose solitudini, permettendone lo sviluppo sinergico e l'integrazione in una scuola, capace, della Giusta distanza.

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applied sciences

Article

Poetry in Pandemic: A Multimodal Neuroaesthetic Study on the Emotional Reaction to the Divina Commedia Poem

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Abstract: Poetry elicits emotions, and emotion is a fundamental component of human ontogeny. Although neuroaesthetics is a rapidly developing field of research, few studies focus on poetry, and none address its different modalities of fruition (MOF) of universal cultural heritage works, such as the Divina Commedia (DC) poem. Moreover, alexithymia (AX) resulted in being a psychological risk factor during the COVID-19 pandemic. The present study aims to investigate the emotional response to poetry excerpts from different *cantica* (Inferno, Purgatorio, Paradiso) of DC with the dual objective of assessing the impact of both the structure of the poem and MOF and that of the characteristics of the acting voice in experts and non-experts, also considering AX. Online emotion facial coding biosignal (BS) techniques, self-reported and psychometric measures were applied to 131 literary (LS) and scientific (SS) university students. BS results show that LS globally manifest more JOY than SS in both reading and listening MOF and more FEAR towards Inferno. Furthermore, LS and SS present different results regarding NEUTRAL emotion about acting voice. AX influences listening in NEUTRAL and SURPRISE expressions. DC's structure affects DISGUST and SADNESS during listening, regardless of participant characteristics. PLEASANTNESS varies according to DC's structure and the acting voice, as well as AROUSAL, which is also correlated with AX. Results are discussed in light of recent findings in affective neuroscience and neuroaesthetics, suggesting the critical role of poetry and listening in supporting human emotional processing.

Keywords: neuroaesthetic; facial emotions recognition; poetry; multimodal; voice

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1. Introduction

Emotions are the essence of life, as colours are the essence of painting and notes the foundation of music. There is growing evidence that our human species has expressed itself in evolution and continues to express itself through colour, ornaments, and other symbolic meanings [1]. Moreover, all humans engage aesthetically with different forms of visual representation, music, dance, literature, architecture, and poetry, so much so that "humans are artists by nature, and the history of art begins with that of humanity" [2] (p. 109). We might add, humanity continues with the study of neuroaesthetics.

Neuroaesthetics, in fact, is often conceived as the study of the neural basis of the production and appreciation of artworks [3–8]. In a broader definition, Nadal and Pearce [9] use the term neuroaesthetics to encompass the study of the neural and evolutionary basis of the cognitive and affective processes enacted when an individual adopts an aesthetic or artistic approach to a work of art, a non-art object or a natural phenomenon. Moreover, an aesthetic experience has been defined as a psychological state determined by interaction with an object to which we intend to attribute qualities according to perceptual, cognitive, affective, or cultural criteria [10]. On the affective aspect, Charles Darwin [11] defended the argument that emotional expressions are involved and adaptive and serve an essential communicative function. In science, emotions can be described as the mental states experienced by humans and are associated with feelings and a degree of pleasure or displeasure [12]; moreover, according to Damasio [13] (p. 84), “the term of emotion should be rightfully used to designate a collection of responses triggered from parts of the brain to the body, and from parts of the brain to others parts of the brain, using both neural and humoral routes”. Different theories have been developed to identify, explain, and categorize emotions [14]. According to the discrete emotion model [15], separate neural systems are responsible for different basic emotions: joy, fear, disgust, anger, surprise, sadness, and neutrality that could be recognized cross-culturally [16–18]. The dimensional model suggests that emotion derives from two neurophysiological systems that can be represented in a two-dimensional structure: arousal and valence. The former concerns the subjective perception of perceived energy intensity (high-low), while valence corresponds to the level of perceived positivity/negativity [19].

Language provides an amazingly versatile and potent means to induce emotions in real life [20]. In addition to communicating information, we use language to make each other feel emotions [21]. Literature makes no exception, as it prompts powerful emotions about unreal events [22,23], which is obvious in poetry. Poetry stems from man’s natural tendency to imitate through language, harmony and rhythm. Poetry is, in fact, defined as the art of creating verbal compositions in which sound and rhythm, i.e., the ‘musical’ dimension of language, take on paramount importance. Initially, and for a very long time, poetry was therefore associated with orality, with transmission by voice. As Plato states in the Republic, while painting is made for sight, poetry is born to be spoken and intended for the ears. Dating back some 43000 years, written poetry is the most ancient record of human literature, and the fact that poetry has accompanied human kind over such a long period, suggests a firm grip on human cognition and emotion [24]. In fact, from the beginning to the present, poetry and emotions are inextricably linked [21]. Furthermore, poetry can generally be understood as inherently concerned with expressing and eliciting affective meaning and emotions [25]. Readers often judge a poetic text by the emotions it conveys; however, empirical research still, as only a few discover how poetry elicits emotions, and only over the last decade have the emotional responses to poetry finally come into focus [26]. However, the emotional impact of poetic language and the associated aesthetic pleasure have yet to be widely investigated with the neuroscientific approach [24]. Some studies [27,28] have shown how literary patterns such as metre and rhythm influence emotions, but, as Pitur and Miu point out [21], empirical research still has a lot to discover about how poetry elicits emotions.

A previous study by our research group [29] investigated, focusing precisely on these literary patterns, the cognitive and emotional neurophysiological reaction to the poem Divina Commedia (DC), a pillar of world culture, written by the Tuscan poet Dante Alighieri (1265–1321). Its repetitive structure is based on the number 3:3 hendecasyllables form a triplet of 33 syllables, the 3 *cantica* (Inferno, Purgatorio, Paradiso) each of 33 *canti* add up to 99 *canti* that with a supernumerary as a proem reach together the perfect number 100 [30]. The work’s structural regularity, the density of historical, philosophical, political and psychological themes, and its unquestionable aesthetic value make it a perfect stimulus for scientific investigations in neuroaesthetics.

How poetry elicits emotions depends on several factors. Among them, the different modalities of fruition (written text, listening to recitation) have always been questioned. While on the one hand, “Cognitive Poetics”—where cognition is to do with the mental processes involved in reading, and poetics concerns the craft of literature—is all about reading literature [31] from a historical perspective, poetic practices throughout the world initially occurred in the form of song and musical, multimedia performance [32–34]. Furthermore, poetry was once only spoken; non-literate cultures still recite it (see, e.g., [23,35]).

As Wagner pointed out [36], readers can experience different emotions in response to the same poem and some reading characteristics, such as the reading experience and psychological traits, can also influence emotions [21]. Studies observe that non-expert readers tend to evaluate poems differently than expert readers [37]. Moreover, alexithymia (AX)—a term that refers to the limitation of emotional function (identifying and describing subjective feelings) [38,39]— might also be interpreted as a dispositional tendency not to express emotions [40]. It is worth noting that the main feature of alexithymia is a deficiency in emotional processing, including atypical eye gaze behaviour, abnormal emotion recognition and emotion processing [41–43] and, differences in the way the brain processes emotions conveyed through the voice [44]. Finally, considering the various psychological situations that emerged in pandemics in young people and adults (see, e.g., [45–47]), evidence shows that among psychological dimensions that mediate the relationship between stressors and mental health outcomes during lockdowns for the COVID-19 pandemic, an important role is played by AX [48].

With the development of advanced and affordable sensor technologies, investigations into emotion recognition have become increasingly popular among affective computing researchers, and recently, there has been a rising trend in research to improve emotion recognition systems with the ability to detect, process and respond to people’s emotional states [49].

In the task of emotion detection, various biosignals or physiological signals—those signals that can provide details about the physiological states and their associated dynamics in the body of a human being [50]—can be used to classify emotions. Recently experiments were conducted using electroencephalography (EEG), GSR (galvanic skin response) [51] electrocardiogram (ECG) [52], electromyogram (EMG) [53], pupillometry [54], (see [55,56] for reviews on emotion recognition based on physiological signals) and physical micro-expressions (ME). ME are spontaneous, subtle, and rapid (1/25 to 1/3 s) facial movements reacting to an emotional stimulus [57,58]. The study of ME provides the ability to expose genuine emotions that occur briefly and unintentionally, even when true emotions are deliberately masked [59,60] for a review on ME recognition). Ekman’s research group over the years specified universal facial expressions in terms of the Facial Action Coding System (FACS) [61], describing sets of facial action units (AUs) specific to prototypical expressions [62]. Finally, facial expression bio-signals figure prominently in research on almost every aspect of emotions, including psychophysiology, neural bases, perception, social process, emotion disorder (see [63] for further details) and art appreciation like poetry [24], and music [64].

In light of the scientific evidence on the different neuroaesthetic and cognitive reactions between experts and non-experts in response to different artistic stimuli [29], [65], the cross-modality in neuroaesthetic response [66] and that emotions from different senses interact at multiple levels [67,68], considering that the experimental protocol was conducted during a period of confinement for COVID-19 that did not allow direct contact between people, the present study has two connected and innovative aims:

I To investigate the different emotional responses (assessed using biosignal-based and self-report subjective measures) according to the structure (*cantica*) and the different modalities of fruition (read/listened to) of Dante Alighieri’s Divina Commedia between literature-skilled and non-literature-skilled students and the possible association with the presence of alexithymia during COVID confinement.

II To investigate whether, while listening to Dante's poem, the coded emotions and self-reported perception vary according to the poem's structure, the qualitative characteristics of the acting voice and the listener's characteristics.

2. Materials and Methods

2.1. Experimental Sample

Before the start of the protocol, a sample of 131 healthy university students was contacted on a voluntary basis through personal networks and social media. The required sample size was calculated a priori using G*Power [69] to ensure that the desired level of power and significant results were able to be achieved. Based on the G*Power output, a sample size of n = 32 was required to detect the effect with a power of 95% and a two-sided significance level of 5%. Finally, 84 healthy participants (18 literature students-LS and 66 non-literature students-SS, mean age 25 years) were included in the study; they did not receive any compensation for participating in the research. All participants were notified of the study and provided digital informed consent before participation. Data was handled following standard practices and in compliance with the GDPR and the European Code of Ethics for Research, and the university's ethical committee approved it. The experiment was also performed in accord with the principles outlined in the Declaration of Helsinki of 1975, as revised in 2000. The project identification code is RM11916B5ADDCB0B.

2.2. Stimuli and Experimental Protocol

The protocol's stimuli consisted of emblematic excerpts from three *canti*, each belonging to the three *cantica* of Dante Alighieri's "Divina Commedia" and were chosen by expert Dante academics. The selected *canti* were, respectively, *canto* V from Inferno (Hell), verses (vv.) 127–138; *canto* XXX from Purgatorio (Purgatory), vv. 67–78 and *canto* XXXIII from Paradiso (Paradise), vv. 133–145 (see Appendix A with the original Italian version by Sapegno [70] and English translation by Longfellow [71]).

The chosen verses were proposed in the modality of reading the text on the screen and listening modality through the acting of two professional Italian actors (male and female). The average duration of the recitation was 42.66 s (sec), and that of the reading (assessed through a pre-test on a sample of ten students outside the research group) was 30 s. At the end of each stimulus presentation, participants were asked to indicate via a Visual Analogue Scale (VAS) [72,73] the arousal (in terms of intensity), pleasantness and positive/negative valence (self-reported perception data) perceived during the stimulation. They were also asked to complete a short questionnaire on recognizing the stimuli and their content (self-declared recall) and a psychological questionnaire to measure alexithymia. After receiving details with an online link, participants were also given calibration instructions to ensure they met the technical requirements of the study. See Figure 1 for a synthetic illustration of the protocol.

2.3. Measures

Biosignals data: The online platform Sticky by Tobii for advanced quantitative research "https://www.tobii.com/products/software/online-marketing-research/sticky (accessed on 6 March 2023)" (already used in published studies [74,75], was adopted to measure emotional expression manifested by the participants (FEAR, SADNESS, JOY, SURPRISE, DISGUST, ANGER, and NEUTRAL see [76]) during the stimulation through Sticky by Tobii Emotion Analysis facial expression recognition tool.

Psychological data: Alexithymia was assessed through the 20 items Toronto Alexithymia Scale (TAS 20) [77]). Each item is scored from 1 (strongly disagree) to 5 (strongly agree) for a maximum total of 100, and it includes three subscales: (a) difficulty in identifying feelings (DIF, difficulty in identifying feelings and distinguishing between emotional feelings and the bodily sensations of emotional arousal); (b) difficulty in describing feelings (DCF; difficulty finding words to express feelings to other); (c) EOT (externally oriented style). TAS 20 values were collected for each participant, and scoring was performed according to the published literature.

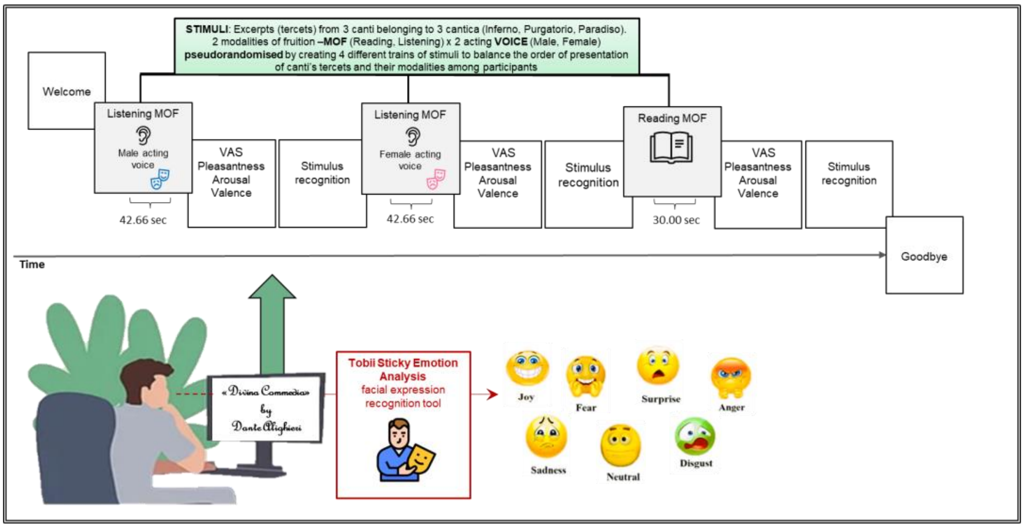

Figure 1. The figure shows the experimental protocol.

Self-reported recall and perception data: each subject's written statement on the recollection of the passages heard and read during the protocol was analysed by authors with experience in literary education to identify whether or not the participant had recognized the verses. Data were collected and analysed together with data on subjective appreciation in terms of perceived pleasantness, arousal and valence.

Online survey Software Qualtrics "https://www.qualtrics.com/it/ (accessed on 8 March 2023)" was used to collect self-reported perception subjective data and psychological alexithymia assessment.

2.4. Statistical Analysis

After checking the normality of each data distribution with the Shapiro–Wilk test [78], independent *t*-tests were used to compare the effects of independent variables EXPERTISE (Literary student (LS); Scientific student (SS) and ALEXITHYMIA (diagnosis of AX or presence of alexithymic traits (AOT)); absence of AX (NA) on dependent variables: seven EMOTIONS (puzzlement, fear, sadness, joy, surprise, neutral, disgust); three SELF-REPORTED PERCEPTION (intensity, pleasantness, positivity) encoded during average listening modality of fruition (MOF) between male voice (M) and female voice (F) and reading MOF.

Subsequently, a factorial analysis of variance (ANOVA) was performed for each facial emotion detected and for each self-reported measure concerning subjective perceptions of pleasantness, intensity (arousal) and positivity (valence). In particular, about poetry pieces considering the factors: CANTICA (three levels: Inferno, Purgatorio, Paradiso), MODALITY of fruition (two levels: listening, reading), VOICE (two levels VM, VF) and GROUPS (LS, SS); (AOT, NA). Moreover, Fisher's exact test [79] was performed on behavioural data (recognition) comparing the two groups (L and S), while Pearson's Chi-squared test (Chi^2 [80] was performed to compare *cantica* (Inferno, Purgatorio, Paradiso). Furthermore, Duncan's post hoc test [81] was used to investigate statistically significant results of ANOVA tests; partial eta squared (η_p^2) [82,83] were computed as measures of the effect size for each dependent variable. Finally, Pearson's Correlation Analysis (r, [84]) was performed to explore the correlation between study variables, while Simple regression analysis was employed to investigate possible directionality between them. *p* values equal to or inferior to 0.05 were considered statistically significant.

3. Results

3.1. How Expertise and Alexithymia Impact Encoded Emotions and Self-Reported Perceptions in Relation to MOF (Listening/Reading) of the Divina Commedia

3.1.1. Impact on Emotions

Concerning the impact of expertise and alexithymia on the emotions detected during the fruition of Divina Commedia proposed in the two MOFs, the results of the *t*-test showed that concerning reading MOF, the LS expressed more JOY than the SS (t = -0.2834, $p = 0.006$ (Figure 2), while in the listening MOF, the AOTs participants expressed more NEUTRAL emotion than the NAs (t = 2.055, $p = 0.043$) (Figure 3).

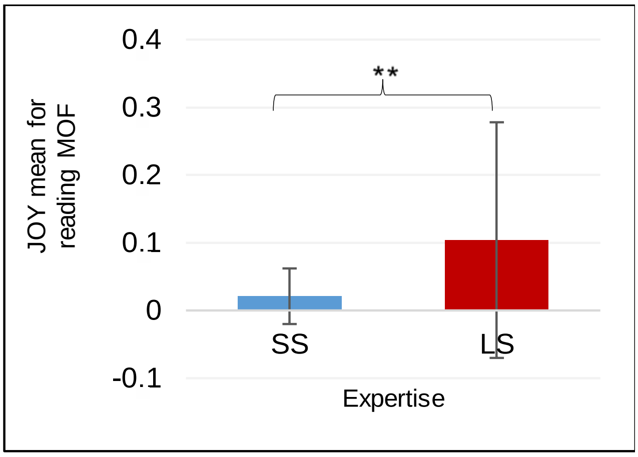

Figure 2. Graph representing the significant difference between Literature (LS) and Scientific (SS) students for JOY emotion resulting from the *t*-test for independent groups analysis. ** $p \leq 0.01$. Bars describe means, and error bars describe standard deviations.

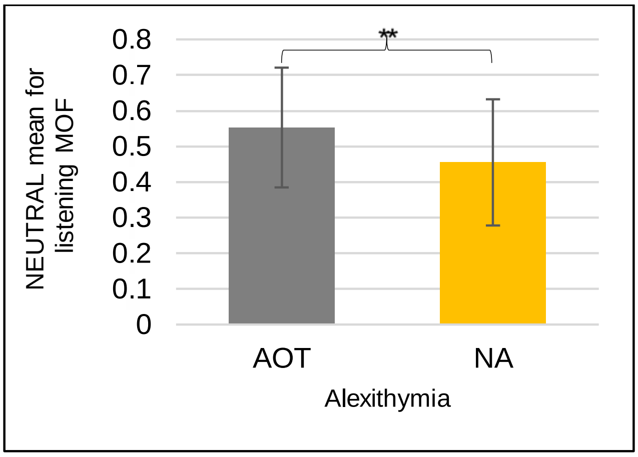

Figure 3. Graph representing the significant difference between participants with alexithymia or alexithymia traits (AOT) and non-alexithymic (NA) students for NEUTRAL emotion during listening modality of fruition (MOF) resulting from the *t*-test for independent groups analysis. ** $p \leq 0.01$. Bars describe means, and error bars describe standard deviations.

3.1.2. Impact on Self-Reported Perception

Concerning subjective perception data about reading MOF, the LS expressed more POSITIVITY than the SS group (t = -2.263, $p = 0.027$, Bonferroni adjusted p value = 0.054) [85] (Figure 4). Furthermore, AOTs perceived more INTENSITY towards the listening modality t =2.589, $p = 0.012$, Bonferroni adjusted p value = 0.036 [85] than NAs (Figure 5).

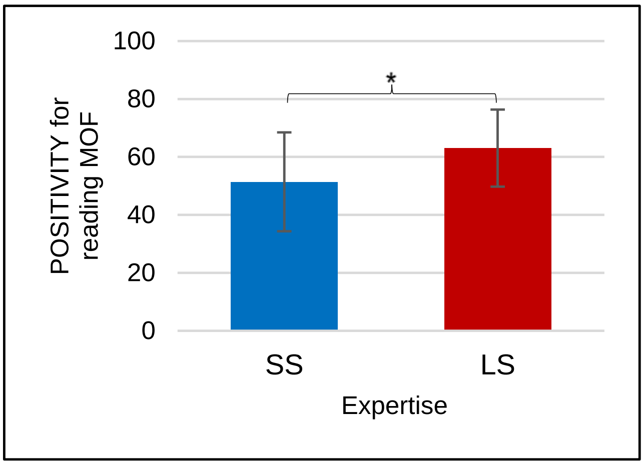

Figure 4. Graph representing the statistically significant difference between literature (LS) and scientific (SS) students for POSITIVITY self-reported subjective perceptions during reading modality of fruition (MOF) resulting from the *t*-test for independent groups analysis. * $p \leq 0.05$. Bars describe means, and error bars describe standard deviations.

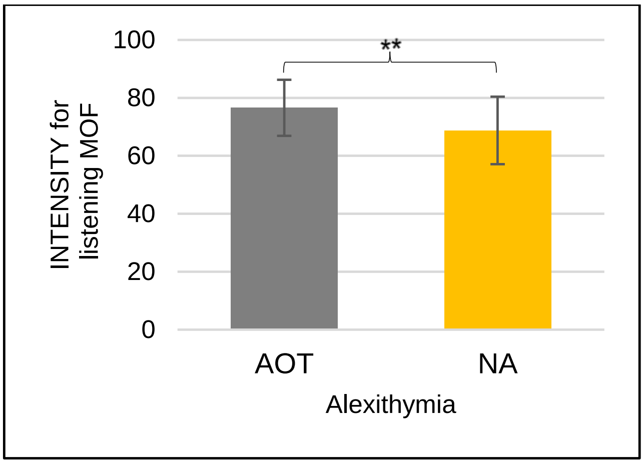

Figure 5. Graph representing the statistically significant difference between participants with alexithymia or alexithymic traits (AOT) and non-alexithymic (NA) for arousal INTENSITY self-reported perception during listening modality of fruition (MOF) resulting from the *t*-test for independent groups analysis. ** $p \leq 0.01$. Bars describe means, and error bars describe standard deviations.

Pearson's correlations analysis showed a positive relationship between the level of alexithymia and both perceived pleasantness (r = 0.28, $p = 0.05$) and intensity (r = 0.39, $p = 0.001$) during the listening MOF. Finally, Simple regression analysis used to determine the causal relationship between independent and dependent variables showed that in our experimental sample, the level of alexithymia is a moderate predictor of the intensity when listening to the divine comedy ($R = 0.329$, $R^2 = 0.108$, $p = 0.012$) (Figure 6).

3.2. How Encoded Emotions and Self-Reported Perception Vary between Groups According to the Structure (Cantica) and the MOF (Listening/Reading) of the Divine Comedy

3.2.1. Emotions Variations

Investigation of interactions between CANTICA × MOF × GROUPS on FEAR by ANOVA analysis revealed a statistically significant interaction among CANTICA and EXPERTISE variables (F (2,72) = 3.868, $p = 0.024$, $\eta_p^2 = 0.097$). Duncan's post hoc showed that, only within the LS group, Inferno revealed statistically significant greater FEAR than Purgatorio ($p = 0.025$) and Paradiso ($p = 0.010$) (Figure 7).

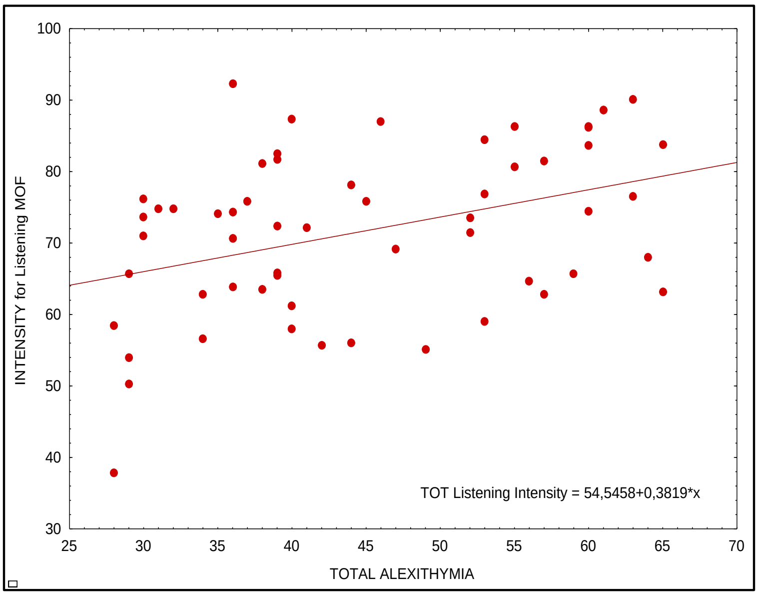

Figure 6. Scatterplot of arousal INTENSITY during listening MOF as predicted by alexithymia.

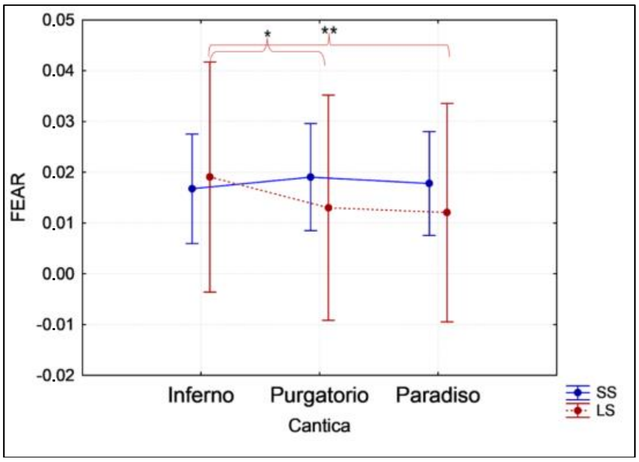

Figure 7. Graph representing the interaction between the factors CANTICA (Inferno, Purgatorio, Paradiso) and EXPERTISE resulting from the ANOVA analysis for FEAR. Vertical bars denote a 0.95 confidence interval. * $p \leq 0.05$; ** $p \leq 0.01$.

Concerning JOY, CANTICA × MOF × EXPERTISE, ANOVA results showed that exposure to listening versus reading MOF elicits greater JOY overall (F (1,24) = 5.6645, $p = 0.025$, $\eta_p^2 = 0.190$) (Figure 8). Duncan's post hoc on the statistically significant interaction in ANOVA among MOF and EXPERTISE variables (F (1,24) = 6.5001, $p = 0.017$, $\eta_p^2 = 0.213$) showed that LS manifest greater JOY for listening MOF in both within and between L/S groups ($0.002 < p < 0.016$) (Figure 9).

3.2.2. Self-Reported Perception Variations

Considering the subjective perception data, ANOVA conducted on PLEASANTNESS showed a statistically significant difference for the CANTICA variable (F (2,110) = 4.200, $p = 0.017$, $\eta_p^2 = 0.070$), from post hoc analysis it emerged that Inferno was considered more pleasant than Purgatorio ($p = 0.001$) (Figure 10).

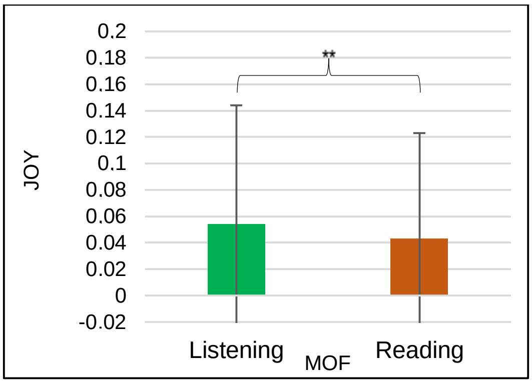

Figure 8. Graph representing the statistically significant difference between listening and reading modality of fruition (MOF) for CANTICA × MOF × EXPERTISE ANOVA for JOY ** $p \leq 0.01$. Bars describe means, and error bars describe standard deviations.

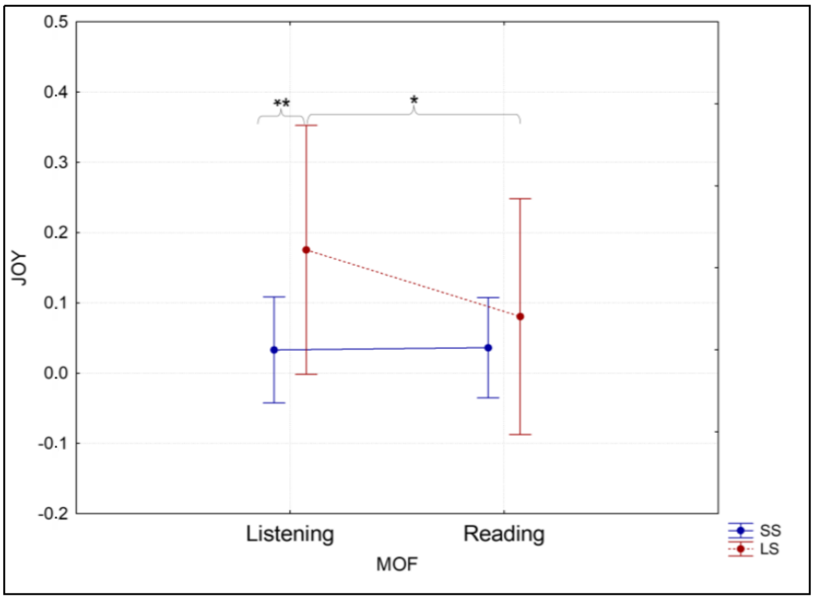

Figure 9. Graph representing the significant interactions among the factors modality of fruition (MOF) and EXPERTISE (Scientific student-SS; Literature student-LS) resulting from the CANTICA × MOF × EXPERTISE ANOVA analysis for JOY. * $p \leq 0.05$; ** $p \leq 0.01$. Vertical bars denote a 0.95 confidence interval.

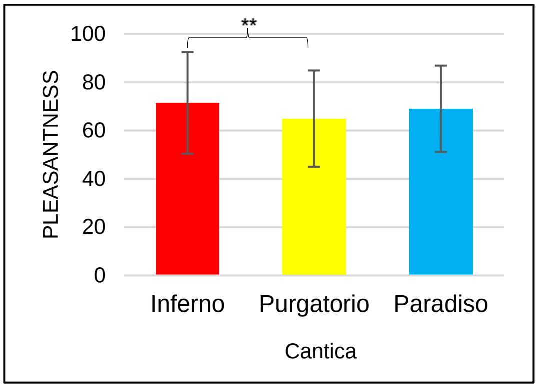

Figure 10. Graph representing the statistically significant difference between the CANTICA variables (Inferno, Purgatorio, Paradiso) resulting from the ANOVA analysis for PLEASANTNESS self-reported perception.** $p \leq 0.01$. Bars describe means, and error bars describe standard deviations.

The ANOVA test concerning POSITIVITY showed a statistically significant interaction among MOF and EXPERTISE (F (1,55) = 4.5897, $p = 0.036$, $\eta_p^2 = 0.077$). Duncan's post hoc revealed that LSs judge more positively valenced the reading than listening MOF both within ($p = 0.023$) and between ($p = 0.042$; $p = 0.017$) SS/LS groups (Figure 11).

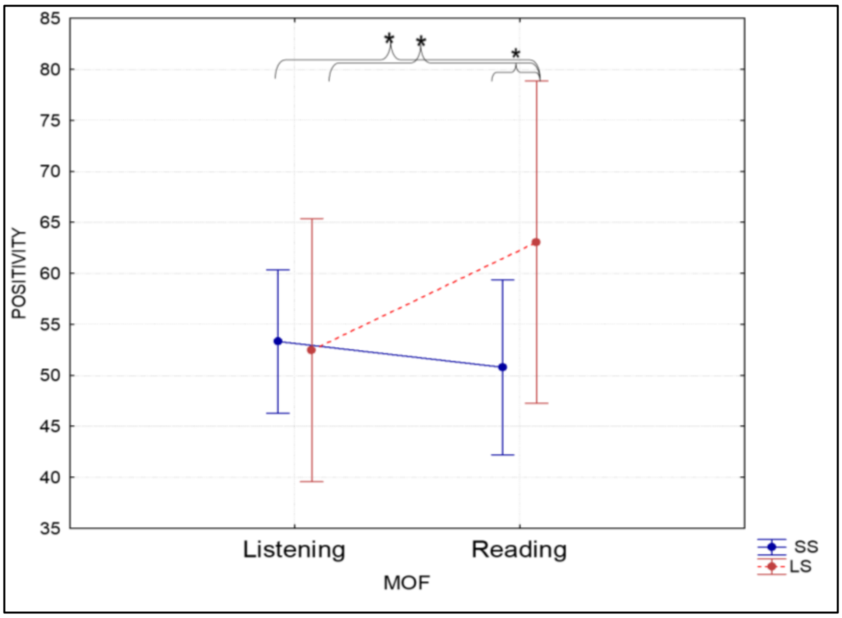

Figure 11. Graph representing the significant interaction among the factors mode of fruition (MOF) and EXPERTISE (Scientific student-SS; Literature student-LS) resulting from the CANTICA × MOF × EXPERTISE ANOVA analysis for valence POSITIVITY self-reported perception. Vertical bars denote a 0.95 confidence interval. * $p \leq 0.05$.

Finally, CANTICA × MOF × group ANOVA for INTENSITY revealed a statistically significant difference for CANTICA (F (2,110) = 4.539, $p = 0.012$, $\eta_p^2 = 0.076$); the Duncan post hoc showed that Paradiso was judged to be less intense than Purgatorio ($p = 0.007$) and Inferno ($p = 0.007$) (Figure 12).

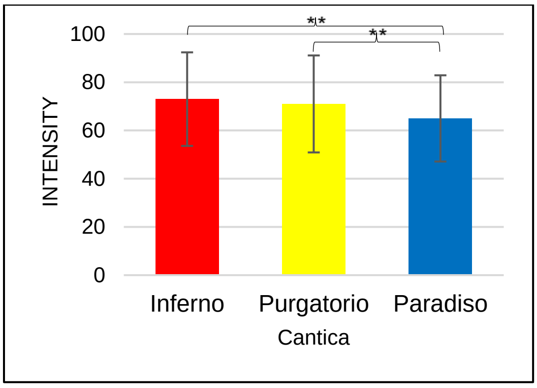

Figure 12. Graph representing the significant difference between the CANTICA variables (Inferno, Purgatorio, Paradiso) resulting from the CANTICA × MOF × EXPERTISE ANOVA analysis for arousal INTENSITY subjective perception. ** $p \leq 0.01$. Bars describe means, and error bars describe standard deviations.

Listening was overall the MOF that conveyed greater arousal intensity to all participants (F (1,55) = 4.874, $p = 0.031$, $\eta_p^2 = 0.081$) (Figure 13).

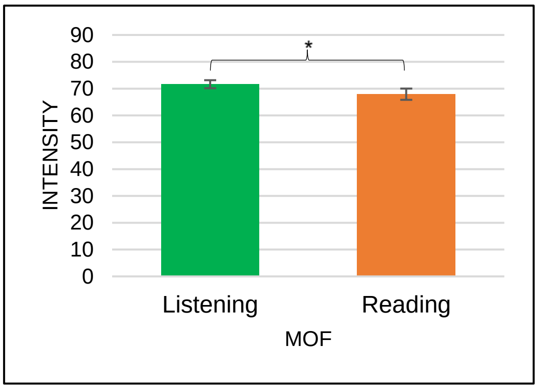

Figure 13. Graph representing the statistically significant difference between modalities of fruition (MOF) resulting from the CANTICA × MOF × EXPERTISE ANOVA analysis for arousal INTENSITY self-reported perception. * $p \leq 0.05$. Bars describe means, and error bars describe standard deviations.

Moreover, the post hoc conducted on ANOVA's significant interaction between MOF × CANTICA (F (2.110) = 4.5011, $p = 0.132$, $\eta_p^2 = 0.075$) showed that listening to Inferno condition, has been perceived to be more intensively arousing than all other conditions (all $p < 0.001$) except for listening to Purgatorio ($p = 0.353$).

3.3. Behavioural Outcomes: Recall

Statistical analysis through Pearson's Chi^2 test, showed a different frequency distribution on the content recall between Cantica: in particular with higher recall rates for Inferno (Chi^2 = 29.80, $p < 0.001$).

3.4. How the Characteristics of the Reciting Voice Influence the Expression of Emotions and Self-Reported Subjective Perception between Groups

3.4.1. Influence on Emotions

The results of the *t*-tests showed that when listening to the Divine Comedy performed by an actor and an actress, AOTs expressed greater SURPRISE compared to NAs for the female voice for Inferno (t = 2.135, $p = 0.038$), Purgatorio (t = 2.094, $p = 0.042$), Paradiso (t = 2.045, $p = 0.046$) (Figure 14).

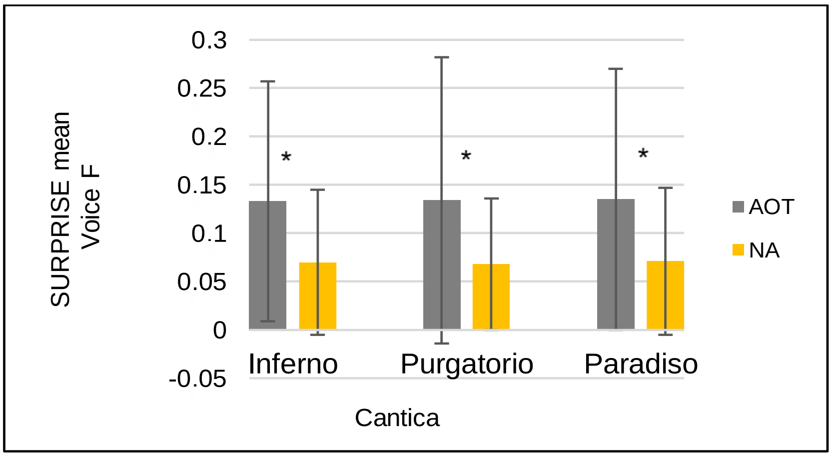

Figure 14. Graph representing the significant differences between participants with alexithymia or alexithymia traits (AOT) and non-alexithymic (NA) for SURPRISE during listening modality of fruition (MOF) for each cantica (Inferno, Purgatorio, Paradiso) resulting from the *t*-tests for independent groups analysis. * $p \leq 0.05$. Bars describe means, and error bars describe standard deviations.

Overall, CANTICA × VOICE × ALEXITHYMIA ANOVA confirmed that AOT perceived statistically significantly more SURPRISE during the overall listening F(1,24) = 7.2531, $p = 0.012$, $\eta_p^2 = 0.232$). (Figure 15).

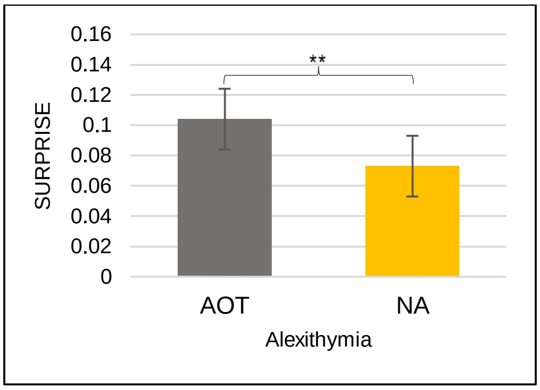

Figure 15. Graph representing the statistically significant difference between alexithymic (AOT) and non-alexithymic (NA) students for SURPRISE emerging from VOICE × CANTICA × ALEXITHYMIA ANOVA analysis. ** $p \leq 0.01$. Bars describe means, and error bars describe standard deviations.

Considering the interaction between CANTICA recited by the two VOICEs and groups, the results of the ANOVA conducted for JOY showed a significant difference between cantica (F (2,48) = 4.251, $p = 0.019$, $\eta_p^2 = 0.150$) and significant interactions among both Cantica × Voice (F (2,48) = 3.270, $p = 0.046$, $\eta_p^2 = 0.119$) and Cantica × Expertise (F (2,48)=3.953, p=0.0257, 0.141). The post hoc on the last interaction shows that Ls manifested significantly more JOY towards Inferno than either Purgatorio ($p = 0.001$) or Paradiso ($p = 0.0024$) (Figure 16).

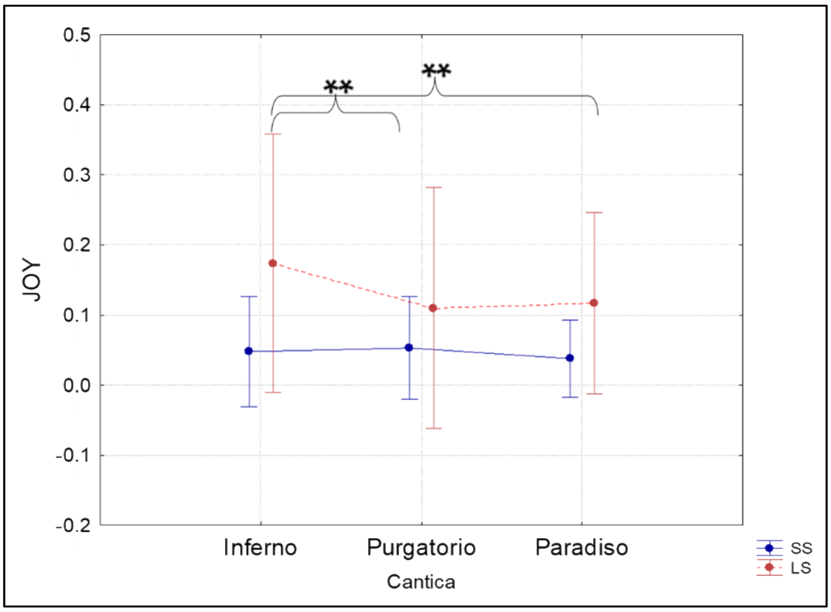

Figure 16. Graph representing the significant interactions among the factors CANTICA (Inferno, Purgatorio, Paradiso) and EXPERTISE (Literature: literature students; Scientific: scientific students) resulting from the CANTICA × VOICE × EXPERTISE ANOVA analysis for JOY. ** $p \leq 0.01$. Vertical bars denote a 0.95 confidence interval.

Regarding NEUTRAL emotion, ANOVA results showed a statistically significant interaction among CANTICA × VOICE × EXPERTISE, (F (2,48) = 3.377, $p = 0.044$, $\eta_p^2 = 0.121$). Duncan's post hoc results (significant only within the L group) are shown in Figure 17.

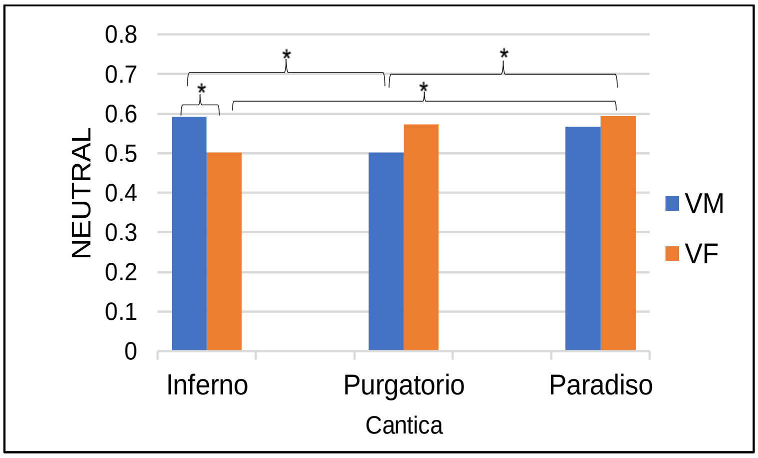

Figure 17. Graph representing the within L students group Duncan's Post hoc results calculated on the statistically significant interaction among the factors CANTICA (Inferno, Purgatorio, Paradiso), VOICE (F: Female; M: Male) and EXPERTISE (L: Literature students; S: Scientific students) resulting from the ANOVA analysis for NEUTRAL. * $p \leq 0.05$; Bars describe means, and error bars describe standard deviations.

Concerning DISGUST, the ANOVA performed on the CANTICA × VOICE × EXPERTISE showed a statistically significant difference between Cantica (F (2,48) = 4.289, $p = 0.019$, $\eta_p^2 = 0.151$). Duncan's Post hoc revealed that Purgatorio is more disgusting than Paradiso regardless of the group ($p = 0.039$) (Figure 18).

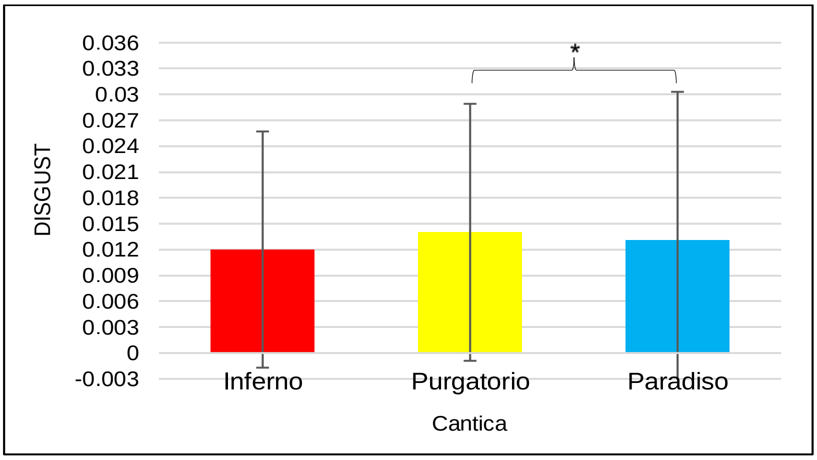

Figure 18. Graph representing the statistically significant difference among the levels of the variable CANTICA (Inferno, Purgatorio, Paradiso) resulting from the CANTICA × VOICE × EXPERTISE ANOVA analysis for DISGUST. * $p \leq 0.05$. Bars describe means, and error bars describe standard deviations.

Concerning SADNESS, the CANTICA × VOICE × ALEXITHYMIA showed a statistically significant difference between cantica (F (2,48) = 5.439, $p = 0.007$). Post hoc showed that overall, Inferno is less sad than Purgatorio ($p = 0.041$) and Paradiso ($p < 0.001$) (Figure 19).

3.4.2. Influence on Self-Reported Perception

Concerning subjective perception data, the ANOVA CANTICA × VOICE × EXPERTISE results for PLEASANTNESS showed a statistically significant difference between the cantica factor (F (2,110) = 5.053, $p = 0.007$, $\eta_p^2 = 0.084$). Duncan's post hoc revealed that Inferno was more liked than Purgatorio ($p = 0.009$) (Figure 20).

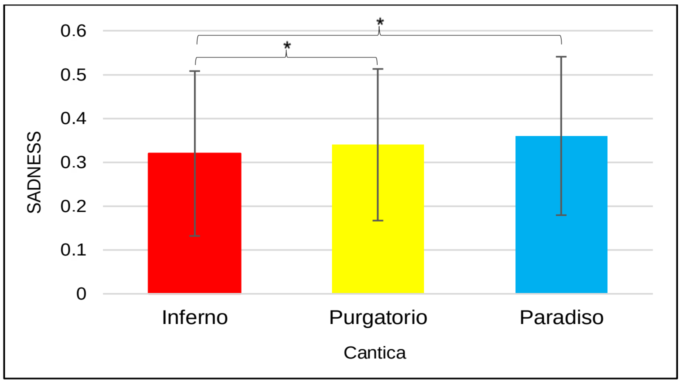

Figure 19. Graph representing the statistically significant differences among the levels of the variable CANTICA (Inferno, Purgatorio, Paradiso) resulting from the CANTICA × VOICE × ALEXITHYMIA ANOVA analysis for SADNESS. * $p \leq 0.05$. Bars describe means, and error bars describe standard deviations.

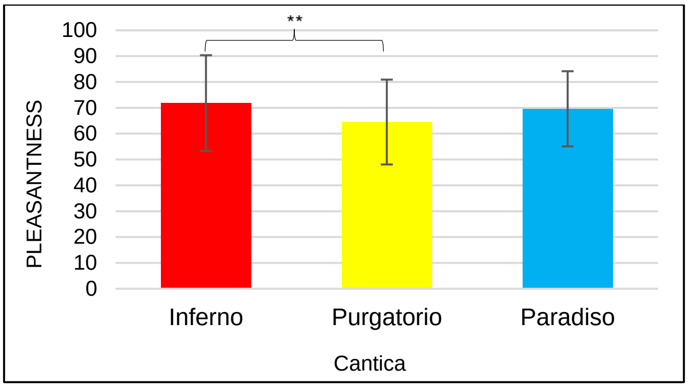

Figure 20. Graph representing the statistically significant difference between variable CANTICA (Inferno, Purgatorio, Paradiso) resulting from the CANTICA × VOICE × EXPERTISE ANOVA analysis for PLEASANTNESS self-reported perception. ** $p \leq 0.01$. Bars describe means, and error bars describe standard deviations.

On the other hand, the post hoc performed on the significant CANTICA × VOICE interaction (F (2,110) = 7.8006, $p = 0.0006$, $\eta_p^2 = 0.124$) showed that Purgatorio recited by a female voice is rated less pleasant compared to all the other conditions ($0.001 > p < 0.02$). At the same time, Paradiso was preferred when recited by a female voice ($p = 0.048$) compared to a male voice. Inferno appeared the only cantica that did not differentiate pleasantness in listening according to the gender of the narrating voice ($p = 0.933$) (Figure 21).

Regarding the perceived arousal INTENSITY, the CANTICA × VOICE × EXPERTISE ANOVA showed that the female voice is overall perceived statistically significantly more intensely than the male voice (F (1,55) = 31.514, $p < 0.0001$, $\eta_p^2 = 0.364$) (Figure 22 left).

Moreover, post hoc calculated on the statistically significant difference between Cantica (F (2,110) = 13.164, $p < 0.0001$) showed that Paradiso is perceived as less intense than other cantica ($p < 0.0001$, $\eta_p^2 = 0.193$) (Figure 22 right).

While post hoc conducted on CANTICA × VOICE significant interactions (F (2,110) = 8.979, $p = 0.0002$, $\eta_p^2 = 0.140$) showed that globally, Inferno recited by a female voice is perceived statistically more intensely than the other cantica and voices ($p < 0.001$). On the other hand, Paradiso recited by a male voice was perceived statistically less intensely than the different cantica–voice combinations ($p < 0.001$).

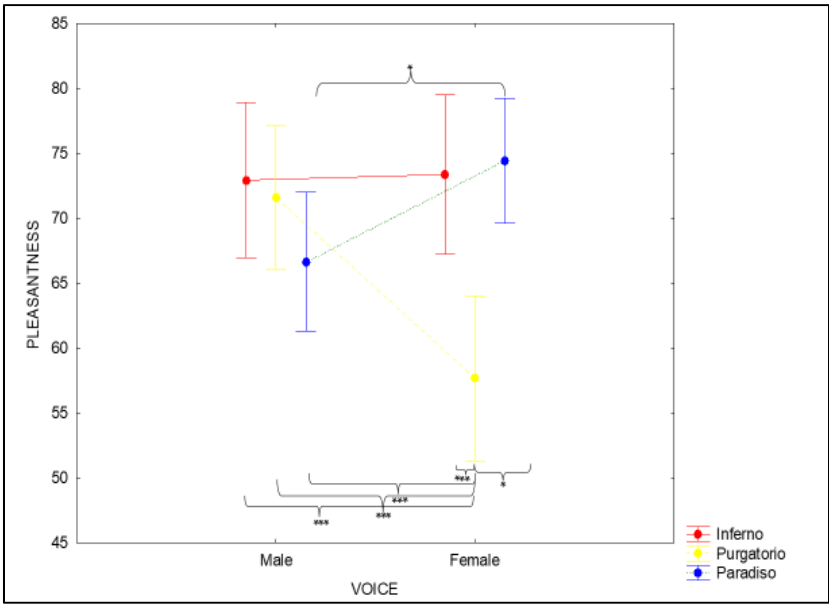

Figure 21. Graph representing the statistically significant interaction among the factors CANTICA (Inferno, Purgatorio, Paradiso), VOICE (Female; Male) resulting from the CANTICA × VOICE × EXPERTISE ANOVA analysis for PLEASANTNESS self-reported perception. * $p \leq 0.05$; *** $p \leq 0.001$. Vertical bars denote a 0.95 confidence interval.

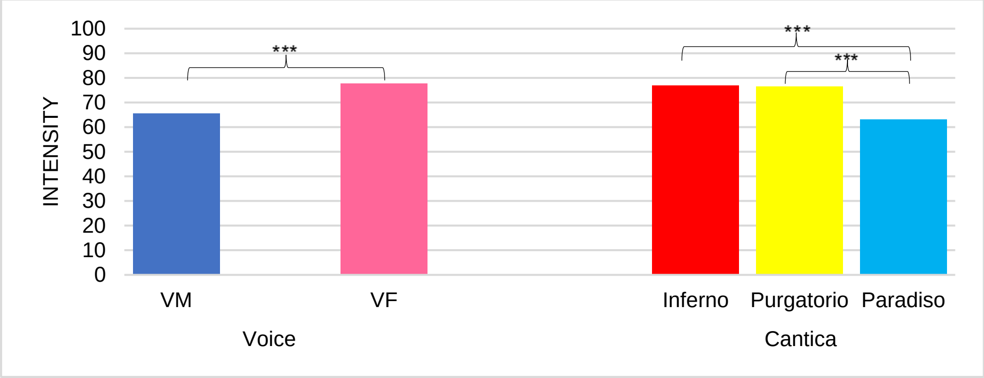

Figure 22. Graph representing results from the CANTICA × VOICE × EXPERTISE ANOVA analysis for arousal INTENSITY self-reported perception. On the left, the statistically significant difference between variable VOICE (VM: Voice Male; VF: Voice Female); on the right, the significant differences between variable CANTICA(Inferno, Purgatorio, Paradiso) *** $p \leq 0.001$. Bars describe means, and error bars represent standard deviations.

4. Discussion

Observing the general comparison according to expertise, it becomes clear that LS manifest more JOY and POSITIVITY towards the reading MOF than SS (Figures 2 and 4). This could be due to the generally greater familiarity with the artistic stimulus (Divina Commedia) in this group based on the expertise, which could lead to less detachment towards the text as suggested from a purely literary perspective by Nabokov [86], who observes that “Curiously enough, one cannot read a book; one can only re-read it. A good reader, a major reader, and an active and creative reader is a re-reader. I shall tell you why. When we read a book for the first time, the very process of laboriously moving our eyes from left to right, line after line, page after page, this complicated physical work upon the book, this stands between us and artistic appreciation”. From a cognitive perspective, Nabokov’s interpretation is ideally in line with the ‘mere exposure effect’ whereby repeated exposure to a stimulus results in more favourable evaluations [87,88], probably due to the greater fluency the perceiver can process an object [89]. To address this possible explanation of expertise effects in our biosignal data for positive emotion (JOY) and positive valence (POSITIVITY), we can refer to the study of Winkielman and Cacioppo [90]. These researchers assessed participants' affective responses to fluent stimuli with facial electromyography (EMG). The EMG is based on the observation that positive affective responses increase activity in the region of the zygomaticus major ('smile muscle'), while negative affective responses increase activity in the area of the corrugator supercilium ('frown muscle'), e.g., [91,92]. As expected, high fluency was associated with increased activity of the zygomatic region (indicative of a positive effect) but not with the movement of the corrugator region (indicative of a negative affect). Presumably, considering our data, L students perceive the positive affect transmitted by the fluency of processing text as their response to Dante's tercets, resulting in more positive evaluations decoded here as JOY and POSITIVITY. Opposite results and interpretations are found in the study conducted by Leder and colleagues [93] investigating through self-reported measures and facial electromyography (EMG) how art expertise modulates the effect of positive and negative works of art on aesthetic and emotional responses in art and non-art students. Authors observe that expertise in emotional response to aesthetic stimuli favours a mode of detachment that attenuates the impact of emotional content on aesthetic evaluation and its physiological correlates. Cartocci and co-workers [29], in line with Leder, in a study investigating different cognitive and emotional neurophysiological responses via biosensors (EEG, GSR) to listening to extracts from the Divine Comedy in literate and non-literate students, suggest an expertise-specific emotional attenuation in the experts towards exposure to poetry. However, such findings were circumscribed to the listening to the acting voice and not in the processing of reading the text, laying the foundation for the present study.

In the comparison, based on the presence of AX, on the other hand, it emerges that in the most unexpected condition (listening to the Divina Commedia as opposed to reading), the AOT group tended to manifest significantly more NEUTRAL emotion (Figure 3) than the NA group, possibly indicating a lack of emotional response [94]. This could be due to the tendency of alexithymic subjects to use expressive suppression as an emotional regulation strategy, possibly stemming from a lack of understanding of their emotions [95]. Furthermore, the higher perceived arousal INTENSITY during listening compared to NA (Figure 5) would seem to be in line with the theory of hyperarousal in alexithymia, which posits that alexithymia is related to higher tonic levels of sympathetic activity and sympathetic reactivity [96], in practice a higher emotional reactivity [97]. Empirical studies have found support for this hypothesis in the visual [98], olfactory [99] and haptic [100] perception of emotions. Our results could provide a contribution to the hyperarousal theory in the auditory modality. Such a hypothesis was supported by our correlation and regression analysis results (Figure 6), showing that the level of alexithymia during listening may predict the intensity the participant manifests. From an art-therapeutic perspective, we could also hypothesize that listening to poetry can, unlike the text, make the alexithymic subject perceive more intensity, and this poetry pattern can be evaluated positively within an art therapy treatment for alexithymia (for art therapy in alexithymia see [101,102]).

In fact, considering that experiential avoidance may be the mechanism (mediating variable) by which alexithymia influences emotion dysregulation [103], one could structure a sort of exposure therapy intervention [104] on listening to poetry to assess a modulation in both self-reported and neurophysiological response. Or, extracts from the Divina Commedia, could be used as part of both individual and group psychotherapeutic poetry interventions [105,106]. Clearly, this interpretative hypothesis should be confirmed by physiological data on the activation of the sympathetic system during a randomized controlled trial combined with additional psychodiagnostic assessments.

FEAR has been one of the most influential emotions in humanity's history [107] and received more scientific attention than any other emotion [108]. Ethologists define fear as a motivational state aroused by specific stimuli that gives rise to defensive behaviour or escape [109]. Already Darwin, at the end of the 19th century, identified fear by defining it as 'states of mind' that we have inherited from our mammalian ancestors by virtue of having inherited certain characteristics of their nervous system [11]. Five centuries before Darwin, Dante Alighieri drew on at least twenty Italian words and expressions to refer to fear. In the Divina Commedia, the word fear appears, in fact 18 times in Inferno, 9 in Purgatorio and 3 in Paradiso. The frequency of use and its numerology indicate that Dante intentionally chooses fear throughout the poem [110]. Especially in Inferno cantica, therefore, the supreme poet's intent seems to be to trigger fear in the reader, an intent captured and made manifest by L students who show more fear for Inferno than for Purgatorio and Paradiso. In contrast, for S students, no significant differences emerge for this emotion (Figure 7). Following the path of experience, therefore, it is always the in-depth knowledge of the text that allows us to grasp the poet's emotional communicative intent and what he first experienced since, as Rea reports [111], Dante's salvific journey begins precisely through fear. In fact, everything begins with the fear experienced in the forest at the beginning of Inferno. For Dante, recounting his extraordinary otherworldly experience means, before anything else, to relive that anguish (Inf. 1.4-6: "*Ahi quanto a dir qual era' cosa dura/esta selva selvaggia e aspra e forte,/che nel pensier rinova la paura*" (Ah, it is hard to speak of what it was/that savage forest dense and difficult/which even in recall renews my fear). Additionally, we would add, make it come vividly alive for the most passionate readers. Moreover, it is interesting to observe how fear is not modulated by the mode of fruition but only by the structure of the Divina Commedia. This latter aspect does not appear on the other hand, from the results that emerged for the emotion JOY, which is overall manifested in the participants more during the listening than during the reading MOF (Figure 8), whereas, considering the expertise, L students express significantly more JOY towards listening than reading the text both within and between L/S groups (Figure 9).

JOY is often used interchangeably with happiness [112]. Experiences of positive emotion are central to human nature and contribute richly to the quality of people's lives [113,114]. Although JOY appears to be essential to the human condition [115] and despite the apparent importance of JOY, it seems to be the least studied of the positive emotions [116], although research on emotions has grown in recent decades [117]. The latest emotion studied but the first to appear if we consider that some experimental data [118,119] indicate that infants smile—and perhaps feel JOY—in the first few days of life. Moreover, within the first few days of life, human neonates can distinguish between expressions of happiness, sadness, and surprise [120], as Panksepp notes [121], supported by evidence showing that the capacity for human laughter preceded the capacity for speech during the evolution of the brain [122], neural circuits for laughter (and so of joy) exist in very ancient regions of the brain. Moreover, behavioural studies have demonstrated that new-born infants prefer human voices to non-vocal auditory stimuli [123], and when presented with vocal expressions with a range of emotional prosodies (happy, angry, sad, neutral), newborns showed an increase in physiological responses following happy prosody [124]. The evidence described so far would seem to lend support to the ancestral capacity of eliciting greater joy during listening than reading MOF. This hypothesis could be further supported by considering the theory of the oral origin of poetry [32,125]. One might speculate that the greatest joy shown in listening to the Divina Commedia might be due to an innate human emotional reaction, sensitivity to vocal auditory stimuli and the oral transmission present at the origins of Western poetry (e.g., [35]). This sensitivity to vocal interpretation may be more pronounced in Ls than in Ss again because of their previous exposure to the artistic stimulus. One could hypothesize that exposure to the text, and prior knowledge in one sensory modality, may increase neuroaesthetic perception towards the same stimulus expressed in another sensory modality as a kind of non-contingent cross-modal emotional correspondence according to the characteristics of the groups (see for details on cross-modal correspondence [126]). However, this interpretative hypothesis needs further investigation to be confirmed, especially in learning.

Considering the subjective value attributed to the verses, the results obtained for the VALENCE and INTENSITY attributed to Dante's terzas show that expertise only influences the first variable. In fact, the Ls attribute more positive valence to the reading than to the listening mode both within and between groups (Figure 11). It is a predictable result and in line with the JOY expressed in response to the text by Ls, given that joy is considered a specific and distinct positive emotion [115].

Focusing on arousal (intensity), it is interesting to note that this does not seem to depend on experience but on the structure of the work and the MOF. Indeed, Inferno is perceived more intensely than Purgatorio and Paradiso (Figure 12). This could be due to the greater recognition of the *cantica* in all participants and the theme of the proposed verses that vividly describe the tormented love between Lancelot and Guinevere. Furthermore, the significantly higher declared intensity for listening than reading (Figure 13) could result from the predisposition to listen to the human voice, a critical skill for survival and social communication [123,127,128]. Finally, the preference for recited Inferno and the predisposition to listen to the human voice seem to be synthesized in the self-report evaluation. In fact, listening to Inferno is perceived as the most intense concerning the other cantica and MOFs except for recited Purgatorio, where no differences are observed. This could be due to the appearance of Beatrice and the force of her direct exclamation, "*Guardaci ben! Ben son, ben son Beatrice*!" ("Look here! For I am Beatrice, I am!"), and the poet's subsequent shame.

Results, moreover, suggest that Inferno is significantly more PLEASANT than Purgatorio, while there is no difference with Paradiso both in the analysis comparing MOFs and, in the analysis comparing acting voices (Figure 10). This lack of differences with Paradiso could be due precisely to the peculiarity of the last canto of Divina Commedia. Here, it is Dante himself who is unable to understand, who strives to find a rational explanation for the mystery of the incarnation in the divine and tells the reader how he lacks the strength to continue to understand and how divine love has appeased his will to know " … .*l'amor che move il sole e l'altre stelle*" (" … the Love that moves the sun and the other stars"). Perhaps it is precisely the incomprehension that Dante conveys to the reader that makes paradise non-comparable in terms of self-reported perceptions because the reader/listener fully experiences the 'aesthetic trance' [129] of the supreme poet. Moreover, in line with previous work of the group [29], the greater appreciation of Inferno compared to Purgatorio can be read in the light of the theory of fluency in aesthetics that predicts higher liking linked to higher recognition of the stimulus [130].

Let us now focus more on voice acting characteristics' effect on the manifestation of emotions and perception in the experts or alexithymic listeners.

Considering the 7 emotions analysed, it emerges that for JOY, listening to Inferno elicits a more significant reaction than listening to Purgatorio and Inferno within the L group (Figure 16). This result perfectly aligns with the first analysis showing that L manifests significantly more joy for listening than for the text (Figure 9) both within and between expertise groups. These results specifically maintain a pattern of appreciation in listening to the Cantica, showing that listening to Inferno induces greater joy than Purgatorio and Paradiso.

The emotion of DISGUST has frequently been implicated in the social context—it appears to serve as an affective mechanism for tracking instances of negative social value, eliciting revulsion and desires for social distance [131,132], outlined the role of disgust in shrinking the moral circle.

Our results showing that the participants manifest more DISGUST towards Purgatory than towards Paradiso (Figure 18) may be due to a social detachment on the part of the listener from what is /declaimed or written in Purgatorio's terzas. The verses in question concern, once again, Beatrice's improvers to Dante for not recognising her. It almost seems as if the listener, more involved in listening to the inferno where the love story between Lancelot and Guinevere is described, or rapt in the contemplative detachment of Paradiso, becomes disgusted with Dante's attitude towards Beatrice, inhibiting the listener's mentalization of the poet. Mentalizing refers to the processes by which we perceive an agent as possessing a mind [133]. Moreover, is the process by which we make sense of each other and ourselves, implicitly and explicitly, in terms of subjective states and mental processes [134]. Thus, one might hypothesise that in listening to Purgatory, the participants detach themselves from the mental interaction that the Poet manages to create with the work user.

Voice characteristics seem to significantly influence NEUTRAL emotion towards auditory stimulation (Figure 17). In fact, Inferno recited by a female voice is less neutral than the male recitation. This result can be explained considering that it is more common to hear Inferno recited by a male voice "https://www.raiplay.it/collezioni/dantealighieri/divina-commedia/divina-commedia (accessed on 6 March 2023)" and by results showing that the Inferno is overall the most recognized cantica. Furthermore, the results show that the female recitation of Paradiso elicits more neutrality than Inferno's and the male recitation of Purgatorio. It could be assumed that as the canto of Paradiso is the least recognised among the participants (and the least studied in schools), the uncommon female recitation makes it almost cathartic, beyond the identifiable earthly emotions. It is interesting to note that the modulation dynamic of neutrality between voice characteristics and cantica only occurs within the L experts, who, again, seem to be more sensitive to the sensory attributes of the auditory poetic stimulus from the perspective of an acquired sensitivity due to the experience of processing the poem.

Curiously, about the different perceived PLEASANTNESS between the cantica according to the acting voice, Paradiso recited by a female voice appears to be the most appreciated among the participants (Figure 21), whereas Paradiso in relation to the other cantica does not differ if the characteristic of the reciting voice is not considered (Figure 20). This is perhaps due to an unconscious transport of the listener who is accompanied through the cathartic tercets of the last canto by a female voice. Just as Dante, in parallel, has been accompanied and protected throughout his journey by Beatrice, with whom he can overcome his fear of hell until he discovers the beauty of *vero amore* (real love) [135]. It is the female voice (like the maternal voice in growing) that guides Dante/the listener through the adversities of the Divina Commedia/Life (for an extended exploration of the female figure of Beatrice, see [136,137]). Speculating again, as mentioned before, we could say that the less pleasantness found in response to Purgatorio recited by a female voice compared to the male recitation could be due precisely to the direct criticism that the Woman makes to Dante/listener. This interpretation could also be supported by the significantly greater INTENSITY expressed in general towards the female voice (Figure 22 left) and the greater INTENSITY perceived towards the female performance of Inferno and Purgatorio compared to the male interpretations, while considering the recitation in general, without the characteristics of the acting voice, listening to Paradiso verses is the least intense perceived compared to the other verses (Figure 22 right). It seems that the preference towards Inferno, already widely discussed, is intensified by the interpretation of the female voice. We do not find these differences in Purgatorio, where, as mentioned, Beatrice criticizes Dante, and the listener could withdraw from identifying with the protagonist indifferently.

The results also show that, overall, Paradiso elicits more extraordinary SADNESS in the listener than Purgatorio and Inferno (Figure 19). This climax between cantiche could be explained by the listener's lack of understanding of an ethereal, non-concrete and ultimately unattainable reality (if we consider that Inferno and Purgatorio are more descriptive cantiche anyway). It could be argued that this data shows the listener's lack of liking for Paradiso. In fact, intuitively, positive aesthetic evaluation and the emotional classification of artworks as joyful or affectively positive seem very closely related [138]. However, movies, music, and poems with sad, (i.e., affectively negative content) have repeatedly been reported to be highly appreciated aesthetically. Likewise, Oliver and Bartsch [139] (p. 31) suggested that the "experience of appreciation is often thought to be tied more closely with sad than joyful affect." Many, if not most, poems are "sad" in terms of their emotional content; readers do not just cognitively decode the emotional context and decipher the emotional expression of poetry, but apparently also genuinely feel the sadness by way of empathy, emotional contagion, identification, or other means of emotional transfer [53,140]. Notably, a rating study of the perception of sad and joyful music excerpts found a significant positive correlation between perceived sadness and perceived beauty [141]. Finally, what Brattico and colleagues effectively propose [142] (p. 2), "tears and joy might co-occur during music listening", we might also imagine applies to the poetry of the Divina Commedia.

While SURPRISE is a common emotion in everyday life, some fundamental characteristics still need to be clarified [143]. Surprise is an emotion arising from a mismatch between an expectation and what is actually observed or experienced in the environment [144]. An expectation is usually thought of as a mental representation of a stimulus or event aroused by some cue or set of cues that has regularly preceded that stimulus or event in the past [145]. Surprise can be seen as an interruption mechanism [146] and motivating people to pay attention to the unexpected stimulus [143].

Previous research into the neural basis of AX has focused chiefly on processing visual emotional stimuli, such as facial or bodily expressions of emotions or emotional pictures and videos. Surprisingly, the impact of alexithymia on the perception of emotional prosody (the melody of speech) has received little attention despite its importance in conveying emotion through the voice in daily conversation [44]. Moreover, a reduced sensitivity to emotional speech prosody in alexithymia was confirmed in a physiological study, in which we additionally observed that alexithymia did not only affect the explicit but also the implicit perception of emotional prosody qualities [147].

It is widely acknowledged that emotions can be communicated through the prosodic features of speech [148], that is, any nonverbal feature, such as pitch, loudness, or rate. For linguists and non-linguists alike, the pitch is the most intuitive and salient gender difference in the voice [149], and gender differences in the agent form the backdrop for much research in phonetics broadly [150].

The anatomy–physiological sexual dimorphism in the vocal apparatus of females and males [151] results in several acoustical differences between female–male adults speakers' voices and, in particular, the mean fundamental frequency of phonation (F0) [152,153]. F0 represents the oscillatory frequency of the vocal cords expressed in Hertz (Hz) and is closely related to *pitch*, defined as our perception of fundamental frequency [154]. Typically, fundamental frequencies lie roughly in the 80 to 450 Hz range, where males have lower voices than females [155]. Interesting studies on the vocal processing of emotions using acoustic parametric biosignals such as F0, show, anger, fear and happiness have all been linked to a high F0 mean and variability [156]. What has been reported so far could be an interpretative support of our data concerning surprise.

In fact, the results show significant differences exclusively during vocal fruition of the Divina Commedia and related to the presence of alexithymia or alexithymic traits. The specific overall AOT shows greater surprise both during the listening (regardless of voice characteristics) of the verses (Figure 15) and during the female interpretation of all canti than NAs (Figure 14). Considering that the Divina Commedia is narrated in the first person by a *male* (Dante Alighieri) and that male audio and video poem's interpretations are generally more common ("https://www.raiplay.it/collezioni/dantealighieri/divina-commedia/divina-commedia (accessed on 6 March 2023)", hearing a *female* voice decanting the poem could, on average be perceived as a bizarre, unexpected event. However, it could be hypothesized that such an unexpected event elicits surprise only in the AOT group. Contrary to reduced sensitivity to prosodic emotional in alexithymics [147], in our experimental sample the opposite seems to be the case. In fact, NAs do not present any surprise concerning the gender of the acting voice. Two interpretative hypotheses could be put forward:

(i) AOTs show greater attention to the signifier than to the meaning of the poem (ii) listening to Dante's poem, especially if interpreted by a female voice, succeeds in distracting alexithymics from the emotional detachment and reduced capacities for emotionalising they may manifest [157,158], offering a sort of art-therapy for emotional disorders.

Clearly, all these interpretative hypotheses should be tested in specific clinical studies. However, these results suggest that listening to poetry affects individuals with deficits in processing and expressing emotions differently.

5. Conclusions and Limits

Based on the experimental objectives outlined at the end of the introduction, the conclusions are summarised below:

I The comparison between MOF and the structure of the Divina Commedia concerning the groups (LS-SS; AOT-NA) shows how the participants' expertise influences the emotions of JOY and FEAR: LSs show more JOY and FEAR towards the poem. Whereas AOTs express more NEUTRALITY towards listening than NAs. In general, listening to MOF is considered more INTENSE, while PLEASANTNESS is not influenced by the mode but by the structure of the work. POSITIVITY is modulated by expertise, while arousal INTENSITY is by the alexithymic factor.

II Considering only auditory stimulation and voice characteristics, besides the greater JOY for listening and NEUTRALITY towards the male voice expressed by LSs, SURPRISE is modulated by alexithymia, whereas the structure of the poem modulates DISGUST and SADNESS. Subjectively, when listening, the female acting voice is perceived as more PLEASANT as well as being considered of greatest arousal INTENSITY.

Our study shows how expertise and difficulties in processing emotions play an important role in the enjoyment of poetic art, suggesting that prior knowledge of the artistic work enables a deeper emotional experience with it by assuming learning support. On the other hand, listening to poetry seems to be capable of vibrating the soul strings of subjects with alexithymic traits, offering hints for possible art-therapeutic paths.

Although the work offers significant results, they should be further investigated on a larger sample and through physiological techniques such as electroencephalography and HR-GSR combination, accompanied by additional psychodiagnostic tests.

Concluding, the present study, unique in its use of modern emotional facial recognition technology, demonstrates the emotional impact of ancient and universal poetry on current students during complicated times such as the pandemic.

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Institutional Review Board Statement: The study was conducted according to the guidelines of the Declaration of Helsinki and approved by the Institutional Ethics Committee of Sapienza University (protocol code RM11916B5ADDCB0B).

Informed Consent Statement: Informed consent was obtained from all subjects involved in the study.

Data Availability Statement: The data presented in this study are available on request from the corresponding author without undue reservation.

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Conflicts of Interest: The authors declare no conflict of interest.

Appendix A

Inferno *canto* V Excerpts (vv. 127–138)	English Translation
Noi leggiavamo un giorno per diletto	One day we reading were for our delight
di Lanciallotto come amor lo strinse:	Of Launcelot, how Love did him enthrall.
soli eravamo e sanza alcun sospetto	Alone we were and without any fear.
Per più fiate li occhi ci sospinse	Full many a time our eyes together drew
quella lettura, e scolorocci il viso;	That reading, and drove the color from our faces;
ma solo un punto fu quel che ci vinse	But one point only was it that o'ercame us.
Quando leggemmo il disiato riso	When as we read of the much longed-for smile
esser baciato da cotanto amante,	Being by such a noble lover kissed,
questi, che mai da me non fia diviso	This one, who ne'er from me shall be divided
la bocca mi baciò tutto tremante.	Kissed me upon the mouth all palpitating.
Galeotto fu il libro e chi lo scrisse:	Galeotto was the book and he who wrote it.
quel giorno più non vi leggemmo avante	That day no farther did we read therein.
Purgatorio *canto* XXX excerpts (vv. 67–78)	**English translation**
Tutto che 'l vel che le scendea di testa,	Although the veil, that from her head descended,
cerchiato delle fronde di Minerva,	Encircled with the foliage of Minerva,
non la lasciasse parer manifesta,	Did not permit her to appear distinctly,
regalmente nell'atto ancor proterva	In attitude still royally majestic
continuò come colui che dice	Continued she, like unto one who speaks,
e 'l più caldo parlar dietro reserva:	And keeps his warmest utterance in reserve:
-Guardaci ben! Ben son, ben son Beatrice.	"Look at me well; in sooth I'm Beatrice!
Come degnasti d'accedere al monte?	How didst thou deign to come unto the Mountain?
non sapei tu che qui è l'uom felice?-	Didst thou not know that man is happy here?"
Li occhi mi cadder giù nel chiaro fonte;	Mine eyes fell downward into the clear fountain,
ma veggendomi in esso, i trassi all'erba,	But, seeing myself therein, I sought the grass,
tanta vergogna mi gravò la fronte.	So great a shame did weigh my forehead down
Paradiso *canto* XXXIII excerpts (vv. 133–145)	**English translation**
Qual è 'l geomètra che tutto s'affige	As the geometrician, who endeavours
per misurar lo cerchio, e non ritrova,	To square the circle, and discovers not,
pensando, quel principio ond'elli indige,	By taking thought, the principle he wants,
tal era io a quella vista nova:	Even such was I at that new apparition;
veder volea come si convenne	I wished to see how the image to the circle
l'imago al cerchio e come vi s'indova;	Conformed itself, and how it there finds place;
ma non eran da ciò le proprie penne:	But my own wings were not enough for this,
se non che la mia mente fu percossa	Had it not been that then my mind there smote
da un fulgore in che sua voglia venne.	A flash of lightning, wherein came its wish.
All'alta fantasia qui mancò possa;	Here vigour failed the lofty fantasy:
ma già volgeva il mio disio e 'l velle,	But now was turning my desire and will,
sì come rota ch'igualmente è mossa,	Even as a wheel that equally is moved,
l'amor che move il sole e l'altre stelle.	The Love which moves the sun and the other stars.

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CONCLUSIONS

The main objective of this thesis, which I pursued during my Doctoral research work, was to understand whether a neuroimaging technique such as electroencephalography (EEG) could provide significantly valid quantitative support for identifying neurophysiological correlates in cognitive and emotional tasks in cochlear implant users and non-users. Actually, the significant results obtained from the various experimental EEG studies presented show how it is indeed possible to obtain concrete explanations for the differences in verbal working memory processing found in psychological and logopaedic assessments. This is an evidence of the impact of multidisciplinarity in this field of research.

In addition, also in the study of emotions, the EEG has proved to be a valuable tool for measuring the difficulties present in hearing-impaired children and adults in the processing of auditory emotional stimuli. This was demonstrated even in the presence of noise, offering a determined and quantitative key to a variable, the emotion, which is often considered aleatory. Moreover, the emotional neurometric indices obtained from the analysis of the EEG signal have been proved significant also in their application to other clinical samples, such as people with substance addiction, in investigating and quantifying emotional aspects often concealed in behaviour.

The main impact of EEG has been in clinical diagnostic. The advancement of medical and engineering research coupled with the clinician's skill has led to the new frontier of cochlear implants. The results presented in this thesis explore a new frontier and constitute a further advancement to diagnostic support and rehabilitation in children and adults with sensory deficits such as hearing loss, identifying EEG as an *open window* into brain mechanisms in this clinical population.

The studies carried out during the pandemic were important because allowed me to observe, isolated students in quarantine beyond *closed windows*. And these investigations, which could not make use of the EEG but of remote methodologies (online questionnaires, emotional facial coding), showed that the global multisensory deprivation experienced did not have significantly different effects on psychological well-being. The presence or absence of deafness unexpectedly highlighted an extreme resilience in the hearing-impaired children and their families. Art also showed scientific value in quarantine. Among the results that emerged, listening to poetry, rather than reading, would seem to be a positive support for students with difficulties processing and expressing emotions.

The path followed in this thesis included many disciplines. Starting from anatomy to clinical and diagnostic evidence in audiology and otolaryngology and psychological evaluations of cognitive and emotional processes. Thanks to the implementation of advanced physical-mathematical analysis techniques used by neurosciences such as the EEG applied to cognitive and emotional tasks, it was possible to offer a quantitative 'oscillating' synthesis to the complexity of the issues addressed by the different disciplinary perspectives. The electroencephalographic waves provide hence the synthesis, the 'basic chords', on which the complexity of the human being is built. The discovery and study of such chords have allowed me to quantitatively understand cognitive and emotional mechanisms in people with and without hearing deficits, thanks to a multidisciplinary working synergy.

FUTURE DIRECTIONS

This PhD thesis provides a strong impetus for the interdisciplinary use of electroencephalography. Therefore, the evidence found in the clinical samples of hearing-impaired children concerning WM and emotions and the psycho-cognitive evidence found for attention prompts me to investigate how these abilities are used in people-to-people interaction. Therefore, one aspect I would like to investigate soon, is the cooperation between normal and hearing-impaired children, also focusing on possible neurophysiological correlates of ‘theory of mind’, a deficient aspect in this clinical sample but fundamental for the healthy construction of relationships with others. To this end, I would like to use hyper-scanning EEG to simultaneously acquire EEG signal from two brains while performing a cooperative task. A further field that I am preparing to investigate, driven by the latest studies on poetry and by ancient auditory passions, is that of musical neuroaesthetics, deepening the analysis of the neurophysiological responses during listening in symphonic music works with the idea of flanking electroencephalography with magneto-encephalography. Finally, the effectiveness of the EEG in patients with cochlear implants to investigate the difficulty of listening in noise will certainly be a push towards its application to further hearing pathologies, such as tinnitus, where the background noise seems to be produced by the auditory system itself.

In conclusion, the prow of my sailboat will turn towards new horizons, hoping to be soon able to write about my exploration along new routes. All that remains for me is to raise the sail and ride the waves, confident that I am sailing on a solid ship with excellent and passionate shipmates at my side.

“But I put forth on the high open sea
With one sole ship, and that small company
By which I never had deserted been”

Dante Alighieri
Divina Commedia, Inferno, canto 26

CONCLUSIONI

L'obiettivo principale della presente tesi, scaturita nel corso del lavoro di ricerca per il conseguimento del Dottorato, è stato quello di comprendere se una tecnologia quale l'elettroencefalografia (EEG) potesse fornire un supporto quantitativo significativamente valido per identificare correlati neurofisiologici in task cognitivi ed emozionali in persone portatori e non di impianti cocleari. I significativi risultati ottenuti dai diversi studi sperimentali presentati dimostrano come effettivamente attraverso *pattern* elettroencefalografici, sia possibile ottenere concrete motivazioni alle differenze nel processamento della memoria di lavoro verbale riscontrabili nelle valutazioni piscologiche e logopediche.
Nello studio delle emozioni, l'EEG ha dimostrato essere un vantaggioso strumento per valutare le difficoltà presenti in bambini ed adulti ipoudenti nel processamento di stimoli emozionali uditivi, anche in presenza di rumore, offrendo una chiave di lettura determinata e quantitativa ad una variabile, l'emozione, spesso considerata aleatoria. Inoltre, gli indici neurometrici emozionali ricavabili dall'analisi del segnale EEG, si sono dimostrati significativi anche nell'applicazione ad altri campioni clinici, come le persone con dipendenza da sostanze, nell'indagare e quantificare aspetti emozionali spesso celati nel comportamento.

Considerando come l'EEG abbia avuto negli anni passati e mostri tutt'ora un forte impatto sulla diagnostica e di come l'avanzamento della ricerca medica ed ingegneristica unite all'abilità del clinico abbiano condotto ad impianti cocleari sempre più di frontiera, i risultati presentati in questa tesi esplorano una nuova frontiera e costituiscono un supporto alla diagnosi ed alla riabilitazione in bambini ed adulti con deficit sensoriali quali l'ipoacusia, identificando l'EEG come una *finestra aperta* sui meccanismi cerebrali in questa popolazione clinica.
Gli studi svolti durante la pandemia, mi hanno permesso di osservare studenti isolati in quarantena dietro le loro *finestre chiuse.* E, questo particolare setting sperimentale, che non si è avvalso dell'EEG ma di altre metodologie applicabili da remoto (questionari online, *emotional facial coding*) ha mostrato che la deprivazione multisensoriale globale vissuta, non ha avuto effetti significativamente diversi sul benessere psicologico in base alla presenza o meno di sordità, evidenziando inaspettatamente una estrema resilienza nei ragazzi ipoudenti e le loro famiglie. Anche l'arte ha mostrato valore scientifico in quarantena. Tra i risultati emersi, l'ascolto della poesia, piuttosto che la lettura, sembrerebbe essere un supporto positivo in studenti con difficoltà nel processamento e nell' espressione delle emozioni.

Il viaggio condotto in questa tesi è stato dunque un viaggio attraverso diverse discipline. Dall'anatomia alle evidenze cliniche e diagnostiche in audiologia ed otorinolaringoiatria ed alle valutazioni psicologiche dei processi cognitivi ed emozionali, grazie all'utilizzo di tecniche di analisi fisicomatematiche avanzate utilizzate dalle neuroscienze quali l'EEG applicata a task cognitivi ed emozionali, è stato possibile offrire una sintesi quantitativa 'oscillante' alla complessità delle problematiche affrontate dalle diverse prospettive disciplinari. L'onda elettroencefalografica, pertanto, sembrerebbe fornire la sintesi, gli 'accordi di base', sui quali si costruisce la complessità dell'essere umano. La scoperta e lo studio di tali accordi mi hanno permesso di comprendere quantitativamente meccanismi cognitivi ed emozionali in persone con e senza deficit uditivi, grazie ad una sinergia di lavoro multidisciplinare.

DIREZIONI FUTURE

Questa tesi di Dottorato fornisce una forte spinta all'utilizzo interdisciplinare dell'elettroencefalografia. Pertanto, le evidenze riscontrate nei campioni clinici di bambini ipoudenti a riguardo della memoria di lavoro e delle emozioni, oltre alle evidenze psicocognitive riscontrate per l'attenzione, mi spingono ad approfondire come queste abilità siano utilizzate nell'interazione tra persone. Pertanto, un aspetto che vorrei presto indagare è quello della cooperazione tra bambini normo ed ipoudenti, focalizzando anche su eventuali correlati neurofisiologici della "teoria della mente", aspetto deficitario in questo campione clinico ma fondamentale per la sana costruzione della relazione con l'altro. A tal fine vorrei utilizzare la tecnica *hyperscanning* che mi permetterà di acquisire in contemporanea i segnali EEG da due cervelli nello svolgimento di un compito cooperativo. Un ulteriore ambito che mi appresto ad indagare spinta dai recenti studi sulla poesia e da antiche passioni uditive, è quello della neuroestetica, approfondendo l'analisi delle risposte neurofisiologiche durante l'ascolto di opere sinfoniche con l'idea di affiancare all' elettroencefalografia la magnetoencefalografia. Infine, l'efficacia dell'EEG osservata in pazienti con impianti cocleari per indagare la difficoltà dell'ascolto nel rumore, sarà sicuramente una spinta verso la sua applicazione ad ulteriori patologie uditive, quali l'acufene, dove il rumore di fondo sembra essere prodotto dallo stesso sistema uditivo.

Concludendo, la prua della mia barca a vela si orienterà verso nuovi orizzonti. Sperando di poter presto scrivere dell'esplorazione compiuta lungo nuove rotte, non mi resta che alzare la vela e cavalcare le onde, sicura di navigare su una nave solida con a fianco eccellenti ed appassionati compagni di navigazione.

"ma misi me per l'alto mare aperto…
sol con un legno e con quella compagna
picciola da la qual non fui deserto"

Dante Alighieri

Divina Commedia, Inferno, canto 26

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FULL LIST OF PUBLICATIONS

1. **Inguscio, B. M. S.,** Cartocci, G., Palmieri, S., Menicocci, S., Vozzi, A., Giorgi, A., ... & Babiloni, F. (2023). Poetry in Pandemic: A Multimodal Neuroaesthetic Study on the Emotional Reaction to the Divina Commedia Poem. Applied Sciences, 13(6), 3720. https://doi.org/10.3390/app13063720

2.Giorgi, A., Menicocci, S., Forte, M., Ferrara, V., Mingione, M., Alaimo Di Loro, P., **Inguscio, B.M.S**... & Cartocci, G. (2023). Virtual and Reality: A Neurophysiological Pilot Study of the Sarcophagus of the Spouses. Brain Sciences, 13(4), 635. doi.org/10.3390/brainsci13040635

3. Giulia Cartocci, **Bianca M.S. Inguscio**, Andrea Giorgi, Alessia Vozzi, Carlo Antonio Leone, Rosa Grassia, Walter Di Nardo, Tiziana Di Cesare, Anna Rita Fetoni, Francesco Freni, Francesco Ciodaro, Francesco Galletti, Lucia Oriella Piccioni, Fabio Babiloni. Music in noise recognition: an EEG study of listening effort in cochlear implant users and normal hearing controls (2023) (Submitted)

4. Nicastri, M., Lo Castro, F., Giallini, I., **Inguscio, B.M.S.**, Mariani, L., Portanova, G., Ruoppolo, G., Orlando, M.P., Mancini, P. Vocal singing skills by cochlear implanted children without formal musical training. Journal of Pediatric Otorhinolaryngology (2023). Journal of Pediatric Otorhinolaryngology, 111605. https://doi.org/10.1016/j.ijporl.2023.111605

5. Valeria Belluscio, Giulia Cartocci, Tommaso Terbojevich, Paolo Di Feo, Marco Ferrari, Valentina Quaresima, **Bianca M.S. Inguscio**, Giuseppe Vannozzi. Facilitating or disturbing? An explorative study to investigate the effect of auditory frequencies on prefrontal cortex activation and postural sway. (2023) (Submitted).

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16. **Bianca M.S. Inguscio**, Maria Nicastri, Ilaria Giallini, Antonio Greco, Fabio babiloni, Patrizia Mancini, Giulia Cartocci. _ Come (si) sentono i ragazzi? Riflessioni su didattica a distanza e sordità ai tempi della pandemia Capitolo in libro: Bambini ed adolescenti a distanza-Il disagio psichico e

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FUNDED PROJECTS

2022 Sapienza University of Rome "Avvio alla Ricerca" for the research project TO OTHERS: neurophysiOlogical evaluation of Theory of Mind and Social Cognition in normal and Hearing loss children during a Cooperative game through HypeRscanning approach) – 3000 euro, 12 months, **Principal Investigator**

2021 Sapienza University of Rome"Avvio alla Ricerca" for the research project WHEMORCHING (Neurophysiological CHaracteristics of an EMOtional auditory/visual WoRkING memory task in Children with and without Hearing loss) – 1500 euro, 12 months, **Principal Investigator**

2021 Sapienza University of Rome Progetti di Ricerca Medi Il ruolo della Memoria di Lavoro nella comprensione del linguaggio nei soggetti adulti portatori di Impianto Cocleare e Protesi Acustica: ruolo del training nelle prestazioni in situazioni di ascolto con rumore – 10000 euro, 12 months, **Group Member**

2020 Sapienza University of Rome Progetti di Ricerca Medi Biomarkers di infiammazione sistemica in pazienti affetti da Sindrome delle Apnee Ostruttiva del Sonno (OSAS): studio prospettico di correlazione tra indicatori di infiammazione sistemica e differenti quadri clinico-terapeutici di OSAS 10000 euro, 12 months, **Group Member**