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The importance of "Scaffolding" in Clinical Approach to Deafness across the Lifespan

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Alla mia famiglia,

a tutti coloro che amo.

Alcuni non ci sono più,

ma il mio amore per loro resterà immutato nel tempo.

Aknowledgements

Thanks to my Professor, Patrizia Mancini; I will always be grateful for the trust placed in me. You taught me a lot beyond academic knowledges: to take risks in order to achieve higher goals; to tolerate imperfections, first of all in myself; not to prevent realization of a project for fear of failing. We have to go and, if needed, we have to fight; this is a precious teaching you gave me.

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Introduction

Affect motivates cognitive flow
much as flames allow torches
to illuminate the darkness
(Jaak Panksepp)

The term scaffolding is basically used in psychology to indicate the help given by one person to another to perform a task. The term comes from the English word “scaffold”, which means something like “providing a support frame”.

The term scaffolding was first used in the psychological field in an article written by Bruner, Wood and Ross, in 1976 and published in the Journal of Child Psychology and Psychiatry. In that article Authors described the ways of interaction between a tutor and a child who has to build a three-dimensional pyramid out of wooden blocks. The term was used as a metaphor to indicate the intervention of a more experienced person who helps a less experienced one to carry out a task, solve a problem or reach a goal that would not be able to achieve without adequate support. A similar and related concept is "Zone of Proximal Development" (ZPD), developed by Lev Vygotsky (1896 – 1934). The ZPD refers to “the distance between the actual developmental level as determined by independent problem solving and the level of potential development as determined through problem-solving under adult guidance, or in collaboration with more capable peers" (Vygotsky, 1978, p. 86). It is, in other words, the difference between what a person can do without help and what can be achieved with a support framework from a more “experienced” partner.

In biology, the term scaffolding refers to the action exerted by some proteins facilitating the interaction of other proteins by increasing signal. The scaffold materials are commonly used for repairing injured tissues, facilitating constructive remodelling while preserving the structural and functional molecular units of the extracellular matrix (Costa et al., 2017).

In audiological field, the “auditory scaffolding” is a key focus to understand the central role of sound in providing a support frame, underlying higher cognitive and emotional development, far beyond the mere linguistic skills (Conway, Pisoni & Kronenberger, 2009).

Finally, in aging studies, scaffolding is a term used referring to those processes present across the lifespan that involve use and development of complementary, alternative neural circuits to achieve a particular cognitive goal. So, the "scaffolding processes" may protect cognitive function in the aging brain, and available evidence suggests that a good cognitive engagement, exercise, social participation may preserve and strengthen the ability to use this mechanism in elderly age (Park & Reuter-Lorenz, 2009).

Throughout the present work of thesis, the concept of scaffolding will be used as a *fil rouge* through the chapters. What I mean for "scaffolding approach", therefore, is nothing more than this: an integrated and multidisciplinary clinical and research methodology to hearing impairments that could take into account persons as a whole; an approach that needs to be continuously adapted and harmonized with the individuals, pursuant to their progress, their limits and resources, in consideration of their audiological, cognitive, emotional, personal, and social characteristics.

In Chapter One some introductory elements related to anatomical and physiological outlines of hearing system, normal and pathological hearing functioning, classification, main etiologies and the potential impact of hearing loss on neuropsychological functioning are provided. Part of the first chapter is focused on cochlear implant, reporting outlines about his functioning, the expanding indications and contraindication and final personal considerations about the importance of some psychological aspects for candidacy.

Chapter Two concerns the emotional, affective, and relational dynamics in families with hearing-impaired children with cochlear implant. The concept of psychological scaffolding will be in turn declined in two different perspectives: as "parental scaffolding" to the child, and as a clinical approach to the family.

From a linguistic and cognitive point of view, parental scaffolding refers to verbal stimulations and input offering children information about objects and actions and age-appropriate problem-solving strategies (Landry, Miller-Loncar, Smith & Swank, 2002). Parental sensitivity and cognitive stimulation turned out to affect language outcomes in children with a cochlear implant as strongly as does age at implantation (Quittner et al., 2013).

Deafness may disrupt mother-child bonding, paving the way for emotional difficulties (Kral & O’Donoghue, 2010): we believe it is essential to take into account the possible intrapersonal and interpersonal dynamics that can be activated when parents receive diagnosis of deafness for their child. At the same time, it is relevant to wonder what the consequences of parental psychological status on their responsiveness might be. Moreover, it is crucial to consider how and to what extent the alteration in natural responsive behaviours and psychological status may deplete not only linguistic but even emotional development of the deaf child.

To offer a psychological scaffolding to parents even before the choice of the proper prosthetic solution, in order to provide a proper time and a proper psychological space for elaboration of difficult emotions is a crucial point in rehabilitation. In the last section of the chapter, a study of our research group designed to assess the effects of parent training (PT) on enhancing children’s communication development will be presented. The PT was based on the “It Takes Two to Talk” model, with specific adaptations for families of deaf children.

The quality of early parent-child interactions made up by verbal scaffolding, autonomy support, sensitivity, and contingent responsiveness has been also positively associated with development of executive functions and emotional development.

In Chapter Three the importance of an early auditory experience for deaf children will be discussed, assuming that the disruption of functioning related to hearing loss need to be conceptualized in a complex framework of physiological, developmental, environmental, emotional and re/habilitative interplaying factor. It is known that brain development bases on dynamic and complex self-organizing processes, including a sequence of neurogenic events. Development of the human connectome - the network map of neuronal connections comprising the nervous system (Petersen & Sporns, 2015) -is strongly linked to interaction between neural activity and environmental sensory experiences (Kral & O'Donoghue, 2010; Hübener &Bonhoeffer, 2014). In case of congenital or very early hearing loss, child could not fully exploit some crucial auditory experiences. According to the “auditory scaffolding hypothesis” (Conway et al., 2009), an early hearing deprivation may deplete cognitive abilities related to learning, recalling, and creating sequential informations, also affecting non-auditory functions related to time and serial order and executive functions. The importance of an early implantation for development of executive functions and emotional competence will be discussed through presentation of two studies of our research group (2016; 2020) concerning variables influencing comprehension of emotions and nuclear executive functions in deaf children with cochlear implant.

In Chapter Four an overview of clinical manifestations of age-related hearing loss is presented, paying particular attention to audiological, psychological, and cognitive issues related to aging with a severe hearing impairment. In old age, biological, cognitive, psychological, and social factors may assume new meanings and different relevance compared to younger ages. Getting older, a reduced ability to discriminate verbal communication in terms of processing speed is one of the issues that occurs most frequently. The hearing-impaired old patient classically presents a discrepancy between the ability to hear sounds and the ability to understand them, especially in noisy environments. Daily and constant hampered and depleted communication may exhaust motivation to communicate, finally impede participation in social life, relational, cultural and aggregation activities. That, in turn, might trigger worsening effects on psychological and /or neuropsychiatric clinical conditions, such as cognitive deterioration, incident dementia and depressive conditions.

Over the past twenty years, research in treatment of severe-profound deafness in adult and elderly patients have increasingly highlighted the need for multimodal and integrated interventions, able to target both audiological and extra- audiological variables related to presbycusis. In other words, it is pointed out the need to extend focus of the intervention beyond the mere correction of the hearing deficit (Kricos, Holmes, & Doyle, 1992; Hickson & Worrall, 2003; Boothroyd, 2007).

In Chapter Five, a revision of existing literature on interventions explicitly focused on elderly hearing-impaired people's psycho-social wellness and quality of life will be presented. The paucity of these kind of interventions and the absence of multidisciplinary rehabilitation protocols for cochlear implant elderly patients was the prompting for building a project named Mind-Active Communication (M-AC) Rehabilitation Program (M-AC; Giallini, Nicastri, Flaccadoro & Mancini, 2020). The "Mind-Active Communication (M-AC) Rehabilitation Program" is intended to be a translation, adaptation, and implementation of Active Communication Education (ACE), a communicative rehabilitation course developed at the University of Queensland, started from the research project of Hickson and Worrall (2003). Original version of ACE program was created to help adults with hearing loss to become more effective communicators, providing them with skills and strategies to cope with everyday difficulties. The Mind-Active Rehabilitation Program is specifically addressed to cochlear implant elderly users affected by severe-profound hearing loss. It is designed to improve metacognitive awareness, problem-solving and self-management in adults with hearing impairment through a multidisciplinary, holistic, and multilevel approach. Last section of chapter five reports a presentation and description of the M-AC program, main topics and aims, multidisciplinary organizations of group and individual sessions with a description of used materials and methodology. Finally, a preliminary evaluation to explore the use of this multidisciplinary rehabilitative program on quality of life, psychological wellbeing, and hearing abilities in a sample of cochlear implanted elderly persons is reported.

I would like to end this introduction by saying that, over these years of research and clinical activity with deaf persons, I gained a growing awareness of the need to adopt an integrated approach in which "*audiological science*" "*brain science*" and "*mind scienc*e" could talk together, providing a support frame, a *scaffolding*, in research, management and rehabilitation of hearing-impaired people. My hope for this dissertation is to convey to readers part of the enriching experience that I have drawn from these years of research and work with hearing impaired people, both from a professional and human point of view.

Clinical studies reported in this thesis were all performed at the Cochlear Implant Center, Policlinico Umberto I - Sapienza University of Rome.

Chapter 1. Hearing Loss and Cochlear Implantation: an overview

1.1 The hearing system: anatomical and physiological outlines

Peripheral hearing system is divided into three portions linearly transducing sounds to nerve impulses: the outer, the middle and the inner ear. When an acoustic stimulus gets the external ear, it is conducted in the middle ear to the inner ear: in this way the auditory processing begins. Vibrations in the air conduct sound to the outer ear: it consists of the auricle and external auditory channel and structurally it is a complex system of cartilage that transmits sound from environment to tympanic membrane. The auricle is a semicircular plate of elastic cartilage with a large parabolic surface and an inherent resonance. These features permit an enhancement of auditory sensitivity by directing sound energy through auditory canal to the tympanic membrane with an amplification of the effective pressure wave.

Vibration of the eardrum transmits sound to the middle ear space where sound travels through vibration of the ossicular system (malleus, incus, and stapes). Malleus is attached to eardrum, whereas the stapes is inserted to the oval window, that is the entry point of the inner ear (cochlea). Although sound pressure waves are conducted to ears by movement of molecules in the air around us, the change from air-conducted waves to movement of fluids in the inner ear makes us able to perceive sounds. When incus vibes, the stapes is driven deeper in the oval window and then retracted, creating a cyclical pushing and pulling on the liquid in inner ear (Alshuaib, Al-Kandari, & Hasan, 2015). Two main physical aspects of middle ear appear relevant in facilitating the transduction process from air-conducted sound wave to fluid movements: first, the surface area of the eardrum is large relative to that of oval window; as a consequence this feature determines a greater pressure transmitted to the cochlear fluids. Second, the malleus is longer than the incus, so vibration of tympanic membrane produces a shorter but stronger movement of the stapes. In normal hearing, the coordinated vibration of the ossicular bones results in a “piston-like” movement of the stapes inserting into the oval window where sounds are transduced into fluid vibrations in the inner ear.

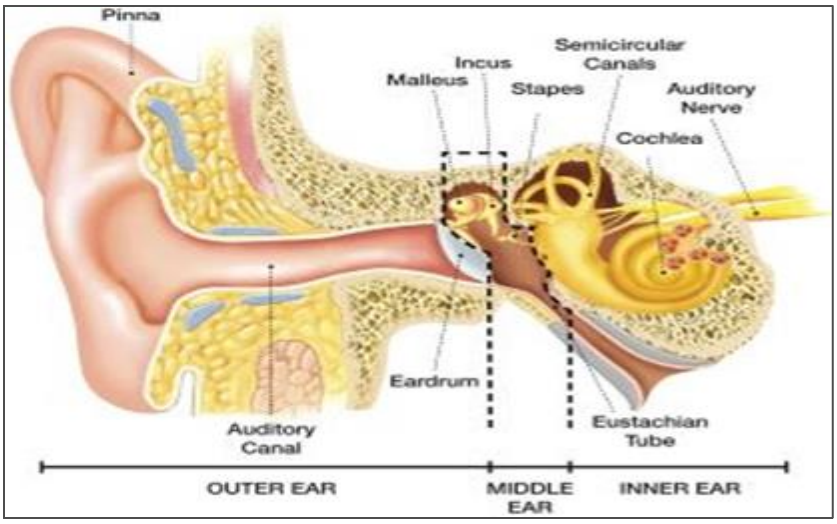
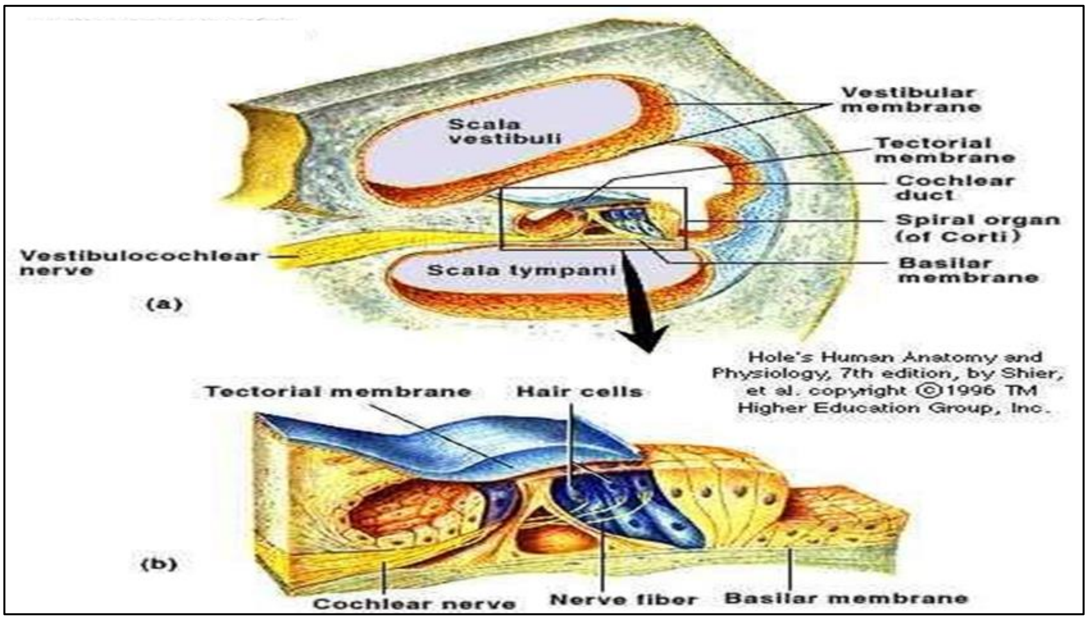


Fig. 1.1 Outer, middle and inner ear

Whereas the middle ear conducts sound to the cochlea, the sensorineural system - cochlea and eight cranial nerve - induces the physiological response to the stimulus, the activation of the nerve cells and the encoding of the sensory response into a neural signal.

The cochlea is divided into three partitions: the scala media or cochlear duct is the cochlear extension of the membranous labyrinth and is filled with endolymph, a fluid rich in potassium (K+) and poor in sodium (Na+). The other two scalae, the scala vestibuli and the scala tympani, are filled with perilymph (with a low potassium concentration) resembling the levels in normal extracellular fluids (Niparko et al., 2000).



Hole's Human Anatomy and Physiology, 7th edition, by Shier, et al. copyright ©1996 TM Higher Education Group, Inc.

Fig. 1.2 Partitions of the cochlea

Fluid vibrations stimulate the auditory receptors cells (hair cells), apically covered with stereocilia, that are attached to the top of basilar membrane within the Organ of Corti. When the cochlea is activated by sound, the scala media and its content - bounded superiorly by Reissner's membrane and inferiorly by the basilar membrane - tend to move as a unit. There are two main types of hair cells: the outer hair cells are nearly 12.000 and are arranged in 3–5 rows along the basilar membrane; the inner hair cells are nearly 3500 and are arranged in a single row. The basilar membrane consists of connective tissue, forming the floor of scala media. This membrane is elastic at the apex of the cochlea, whilst at the base (near the stapes and oval window) it is stiffer: this means that the basilar membrane is more sensitive to low frequencies at the apex and to high frequencies at the base. In other words, when sound gets the ear, it generates a wave that travels from the base towards the apex of the cochlea. A functional cochlea acts as a real spectrum analyser: each place of the cochlea vibrates at his own specific frequency leading a shift of hair cells. Hair cells in the different regions of the cochlea are maximally stimulated by different frequencies. This results in a spatial representation of sound frequency across the basilar membrane where the hair cells are located. Von Békésy (1970) demonstrated that a tone of a certain frequency caused the highest vibration amplitude at a certain point along the basilar membrane. This means that each point along the basilar membrane is tuned to a certain frequency and a frequency scale can be identified along the cochlea, with high frequencies located at the base and low frequencies at the apex of the cochlea (Figure 1.3). As a consequence, each hair cell produces responses that, near the threshold of hearing, are tuned to a characteristic frequency. The result is a tonotopic representation: it is the fundamental principle of organization in the auditory system.

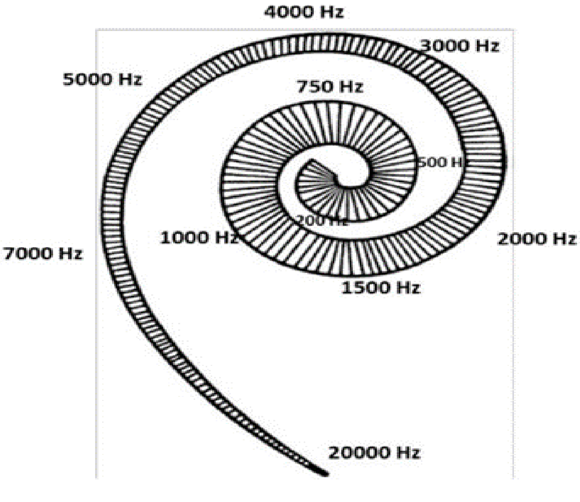


Fig. 1.3 Tonotopic representation of the cochlea

The aspects of the central nervous system that deal with this neurally encoded signal are generally called Central Auditory Nervous System.

Neural signals enter the central auditory system through auditory nerve: translation of sound energy into nerve signals becomes more and more complex and organized when it enters the central auditory system through the auditory nerve to the nuclei of the brainstem.

Midbrain, medulla oblongata and pons are often collectively referred to as the brainstem**,** comparable to the "stem" from which cerebral hemispheres and cerebellum sprout (Figure 1.4).

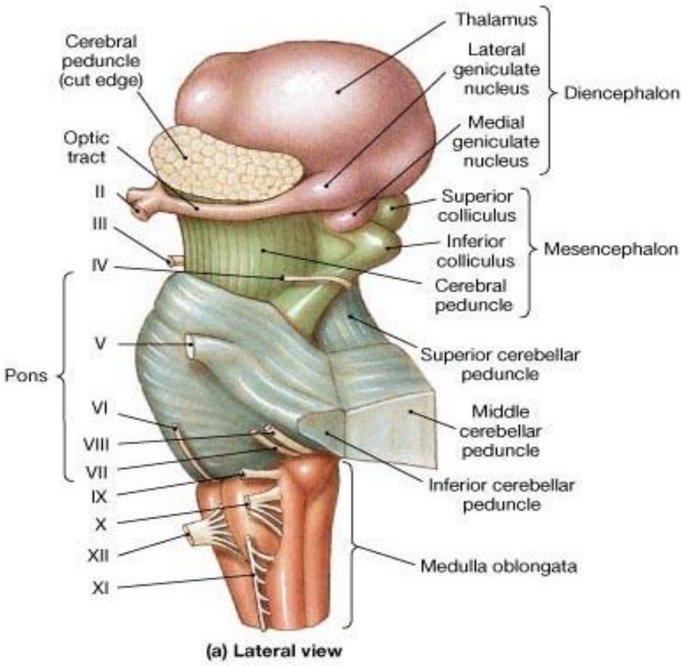


Fig. 1.4 The brainstem

The midbrain is run by cerebral aqueduct (or aqueduct of Sylvius). The dorsal region is called tectum and the ventral region is called tegmentum. The ventrolateral surface of midbrain is located by cerebral peduncles, containing primarily descending fibers including motor commands descending to the spinal cord and sensory information going to the thalamus.

Dorsally, the tectum consists in corpora quadrigemina, two pairs of sensory nuclei containing several areas of gray matter and areas of reticular substance. The superior colliculi process some visual information, aid the decussation of several fibers of the optic nerve (some fibers remain ipsilateral), and are involved with saccadic eye movements. The inferior colliculi - located just above the trochlear nerve – receive auditory information from the medial geniculate nuclei, generating reflexive responses. Ventrally, tegmentum includes the substantia nigra (black substance) consisting of gray matter containing blackish pigmented cells. As part of basal nuclei, substantia nigra plays a role in starting and control of skeletal movement. In the rostral portion of midbrain we found substantia rubra (red nuclei): it is a prominent bloody region playing a role in control of skeleton movements. Medulla oblongata follows the spinal cord and it looks likes an upper base cone trunk made up by white matter bundles, several gray matter nuclei and by reticular substance. Placed transversely in front of the medulla, the pons is located, separated by a furrow. It contains numerous nuclei of gray matter and bundles of white matter and is also crossed by the anterior extension of the reticular substance.

Medulla oblongata includes nuclei that serve as relay stations along sensory and motor pathways and automatic nuclei that include cardiovascular and respiratory rhythmicity centers. At this level also the VIII cranial nerve (auditory nerve) is placed: from an auditory point of view, once sound as mechanical energy is transduced into fluid vibrations in inner ear structures, stimulation of hair cells receptors (stereocilia) occurs, with subsequent synapse with the VIII cranic nerve and entering of information via the brainstem auditory nuclei upon central auditory system. At this level translation of sound energy to neural information becomes more organized and complex, bifurcating into separate monoaural and binaural pathways (see Figure 1.5).

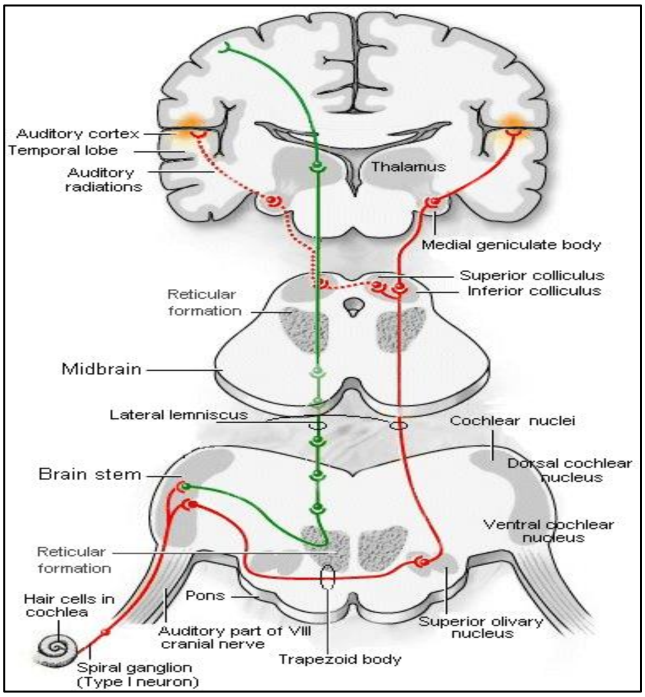


Figure 1.5 The Auditory pathways

The monaural pathway transmits signal amplitude and pitch from each cochlea to the contralateral auditory cortex. The binaural path follows a more complex way, transmitting the signals from each cochlea both ipsilaterally and contralaterally, allowing signals' comparison based on temporal differences and loudness perception, also facilitating discrimination and localization of sound. This is particularly relevant in cases of unilateral hearing loss: since information on pitch and amplitude is sent from each cochlea separately to the auditory cortex, in single-side deafness (SSD) auditory information from an ear is preserved. However, because of SSD, a failure in timing and loudness comparison of the sound occurs, severely hindering correct localization and separation of the sound.

It must be considered that in human auditory system the ears do not work in a perfect symmetrical way; rather, ears respond to different informations then reprocessed and synthesized in a single signal through the complex reception process of binaural hearing. In other words, binaural hearing relies on brain's ability to combine two different sound messages in a third synthetic one. The central binaural interface occurs mainly and almost simultaneously at three levels, which are: - the superior olive complex (SOC), which receives information from both anterior ventral cochlear nuclei (CN); - the nucleus of the lateral lemniscus (NLL), which receives direct projections from the anterior lateral control ventral cochlear nucleus and from the bilateral upper olivary complex; - the inferior colliculus, which receives direct projections from the contralateral ventral cochlear nucleus, from the bilateral superior olivary complex and from the bilateral lateral lemniscus nucleus. Eventually the monoaural and binaural pathways join in the inferior colliculus and are transmitted through the thalamus to the primary auditory cortex. The tonotopic organization is also maintained in the auditory cortex, with the low frequencies being processed rostrally and the high frequencies at the caudal level.

1.2 Classification of hearing loss

Hearing loss (HL) can be classified on the basis of various aspects that firstly and directly impact typology and kind of available interventions. Understanding classification of hearing loss is an essential step for improving preventive approaches, therapeutic management, and adequate rehabilitation protocols.

Concerning physiological parameters, it is to consider the degree and frequencies configuration of hearing loss, the site of disruption in sound transmission and the time of onset. An adequate intervention can never be selected without these essential informations.

Firstly, a classification of HL requires the degree of impairment. Broad categories of hearing loss degrees are represented in Table 1.1. Degree of HL is defined by threshold at which sound is detected across the frequency spectrum. It is measured by amplitude in decibel hearing level (dBHL) across several frequencies (pitch) that are measured in Hertz, or cycles per second. The impact of an HL pattern across the frequency spectrum ranging from 500 to 4000Hz will be more severe than an HL outside these primary speech frequencies. As a matter of fact, audiometric configuration of HL is an essential factor to take into account.

Normal hearing: 0-25 dB	Normal threshold is between 0-25 dB for adults and between 0-15 dB for children.
Mild hearing loss: 25-40 dB	Some difficulties with normal conversation especially in noisy surroundings. Able to hear and repeat words spoken in normal voice at 1 meter. Counselling is recommended and hearing aids may be needed.
Moderate hearing loss: 41-70 dB	Problems hearing in most situations. Able to hear and repeat words spoken in raised voice at 1 meter. Hearing aids and counseling are usually recommended
Severe hearing loss: 71-90 dB	There is no perception of human voice and most of sounds. Without prosthetic intervention, language development is impossible
Profound hearing loss: >90 dB	Deafness: nothing at all can be heard

Table 1.1 Classification of hearing loss

Depending on the site of disruption in the transmission of signal, an HL can be conductive, sensorineural, or mixed, as a combination of conductive and sensorineural damages in the same ear.

Conductive hearing loss (CHL): in CHL external and/or middle ear components present a disruption. In this type of HL, disfunction is in conduction sound from outer ear to eardrum, through ossicles system to the inner ear (Moller, 2006). Generally, CHL results not greater than moderate-severe (41-70dBHL). As sound signals approximately greater than 60 dB vibrate in the skull, effectively bypassing middle ear, CHL is usually associated with better hearing thresholds for bone-conducted rather than air-conducted signals. Common and usually treatable causes of CHL are: ear infections, outer or inner ear inflammation, otitis media, agents acting on transmission of sounds (such as ear wax or foreign object), allergies, problems with Eustachian tube, or trauma injuring eardrum (Kammerer et al., 2010). Other common causes of CHL are atresia and microtia (absence and/or malformation of outer and/or middle ear); conductive pathologies, such as cholesteatoma and tympanosclerosis, the latest often as a consequence of chronic otitis media leading to reduced mobility of eardrum and ossicles system. Usually, CHL affects low and mid-range frequencies (250-2000Hz); nevertheless, in some case all frequencies range can be affected. Common treatment of CHL include use of antibiotics, medication, removal of wax. surgery, amplification with hearing aids and use of assistive devices.

Sensorineural hearing loss (SNHL): SNHL occurs as a result from deficit in the inner ear, commonly a damage of cochlear inner cells in the auditory periphery. Unlike CHL, in SNHL air and bone conduction thresholds are similar. In this type of HL, because the sensory deficit is usually due to a damage to the organ of Corti, the central auditory system is still functional: as a consequence, treatment's goal is to provide the brain with alternative input. When hearing loss is severe-profound (most cases of SNHL), a direct stimulation of auditory nerve with electrical current is the only chance to achieve this goal (Niparko et al., 2000). SNHL can be congenital, prelingually acquired or post lingually acquired; it may occur prenatal (occurring before birth) or perinatal (occurring during birth). A congenital hearing loss can be hereditary and syndromic; hereditary and not syndromic, not hereditary but arising from chromosomic alteration, maternal infections during pregnancy or caused by ototoxic drugs for medical treatments. The developments in molecular medicine made it possible more detailed etiological classification of congenital hearing losses, whose causes were previously indistinguishable (Kral & O'Donoghue, 2010). As a matter of fact, congenital deafness affects several molecular processes which cochlear function relies on.

In addition, there is evidence that genetic variations, such as a mitochondrial mutation, may cause an increased sensitivity to ototoxic agents. Mutation of a single gene (GJB2), which encodes the connexin 26 molecule, is commonly involved in deafness: this kind of mutation causes an interruption in potassium recycling, resulting in the accumulation of potassium and, ultimately, cell death. As descripted above, the inflow of potassium into the hair cells is a fundamental condition for hearing function. In the stria vascularis, potassium is extracted from blood and actively secreted into the endolymph. Deafness may be caused by disturbance of transduction mechanisms, by disturbance in the transport of potassium from hair cells through connexins, or by impairment of its return or secretion into the endolymph (Kral & O'Donoghue, 2010).

In adulthood, hearing loss can be caused by prolonged exposure to loud noise, or sudden exposure to noise at 120-150dB (as, for example, an explosion), autoimmune diseases (such as, for example Meniére disease), ototoxic drugs, trauma, auditory nerve tumors, chronic otitis media. Anyway, HL in adults is most frequently caused by the cumulative effects of aging, so called age-related hearing loss, or presbycusis (Kammerer et al., 2010).

Knowing the cause of hearing loss is essential ~~for~~ an adequate identification, treatment and care of any-age hearing impaired persons. Hearing loss is a severe sensory impairment, but it does not necessarily result in specific neuropsychological, cognitive, intellectual, emotional or social outcomes. Consequences of hearing loss on global functioning depend on a complex interplay of factors that needed to be considered. The impact of even a mild-moderate unaided HL can represent a significant challenge for neuropsychological development, affecting language acquisition, executive functioning, academic outcome, psychological aspects, and the general quality of life.

It should be clear that hearing loss cannot be viewed as a monogram. It can be sudden or progressive, temporary or permanent, bilateral or unilateral, stable or fluctuating. Moreover, a hearing loss can be caused by many factors, such as genetic causes, physical traumas, chemical or illness consequences, prolonged exposure to noise (Kammerer et al., 2010).

Knowing the etiology of a hearing loss is crucial for understanding his potential impact on neuropsychological development, social functioning and quality of life. Anyway, even in case of a similar degree of hearing loss and site of disruption of signal transmission, a child with a congenital hearing loss cannot be considered similar to an older adult with an age-related hearing loss. Furthermore, two children both affected by profound hearing loss, with the same etiology, same frequency spectrum, same site of disruption of signal transmission may have quite different neuropsychological, linguistic, relational, emotional and social trajectories (Kammerer et al., 2010). As it will be discussed later in the present work, the disruption of functioning related to hearing loss need to be conceptualized in a complex framework of physiological, developmental, environmental, emotional and re/habilitative interplaying factors. In Table 1.2, main etiologies of HL and potential impact on neuropsychological functioning are synthetically presented.

Type (Hereditary and Syndromic)	**Etiology**	**HL characteristics**	**Neurological/Neuropsychological correlates**
	Waardenburg syndrome	Range of sensorineural hearing loss from unilateral to bilateral, typically low frequency HL	No known cognitive, psychiatric or neurological implications
	Usher syndrome	Sensorineural hearing loss coupled with vision loss (retinitis pigmentosa), variable age of onset, depending on type (I, II, III)	No known cognitive or psychiatric implications, progressive vision loss; vestibular and gross motor deficits may be involved
	Pendred syndrome	Congenital, non poregressive, severe to profound hearing loss	No known cognitive, psychiatric or neurological implications
	Mitochondrial DNA mutations	Sensorineural, variable range	Cognitive and neurological implications

Type (Hereditary and Non -syndromic)	**Etiology**	**HL characteristics**	**Neurological/Neuropsychological correlates**
	Otosclerosis	Conductive HL, progressive in nature	Social isolation---
	GJB2 gene mutation (Connexin 26)	Account for up 50% of all non-syndromic sensorineural deafness	No cognitive, psychiatric or neurological implications

Type (Non-hereditary) Congenital infections	**Etiology**	**HL characteristics**	**Neurological/Neuropsychological correlates**
	Cytomegalovirus (CMV)	Most common cause of non-hereditary deafness; 40% of CMV neonates develop sensorineural HL	Significant neurological implications, microcephaly, chorioretinitis, selzures, Hypo/Hypertonia, cerebral palsy, mental retardation, behavioural disorders
	Rubella	Exposure to rubella virus in utero can cause sensorineural hearing loss of varying degrees	Possible mental retardation and autistic features; significant physical implications, retinopathy, microcephaly. Micro-ophthalmia, cardiac defects
	Toxoplasmosis, Siphilis, Herpes simplex	sensorineural hearing loss of varying degrees	Cognitive and neurological implications, such as mental

			retardation, visual impairment, seizures

Type (Non-hereditary) Other etiologies	Etiology	HL characteristics	Neurological/Neuropsychological correlates
	Bacterial meningitis	Sensorineural HL	Cognitive, language, behavioural problem; neurological implications such as mental retardation, spasticity, paresis, seizure disorders, specific language deficit
	Auditory nerve tumors	Profound sensorineural loss resulting from noncancerous tumor often arising from vestibular nerve	Balance problems; hemi-facial problems after surgery; difficult emotional adjustment to HL
	Hypoxia	Variable sensorineural HL, often associated with prematurity	Variable outcomes on neurological and cognitive functioning
	Noise-induced HL		
	Ototoxic drugs	Sensorineural, variable in severity and in onset after drug exposure	No neurological implication, but adaptation can be difficult
	Presbycusis	Progressive HL, bilateral, initially on higher frequencies	Often associated with depression, anxiety, isolation, paranoic symptoms, faster cognitive decline (?)
	Otitis media	Conducitve, mild to modearet degree of HL	Mild deficits in working memory and language processing associated with later language and learning disabilities

Table 1.2 Main etiologies of HL and potential impact on neuropsychological functioning (adapted from Kammerer et al., 2010)

1.3 Cochlear implantation: functioning, indications, and contraindication for candidacy

Cochlear Implant (CI) is a sensory neuro-prosthetic device representing "gold standard" in treatment of profound severe hearing loss; it works through an electrical neuronal stimulation able to imitate the natural physiology of sensory organs. When CI is activated, it generates an electrical response, targeting spiral ganglion and auditory nerve fibres, bypassing organ of Corti. The artificial electrical stimulation of CI is interpreted by brain as a sensory auditory input innervating the receptor cells.

Nowadays in the world, more than 80.000 deaf children are using a cochlear implant (Kral & O’Donoghue, 2010) and approximately 330.000 registered devices have been implanted worldwide (National Institute of Health Publication, NIH, 2016).

Cochlear implant (Figure 1.6) consists of two main components:

1) an external device, consisting of microphone, speech processor (elaboration unit), and transmitter coil (transmission unit);

2) an internal device, implanted behind the ear, including the receiver electronics (receiver coil) and an electrode array.

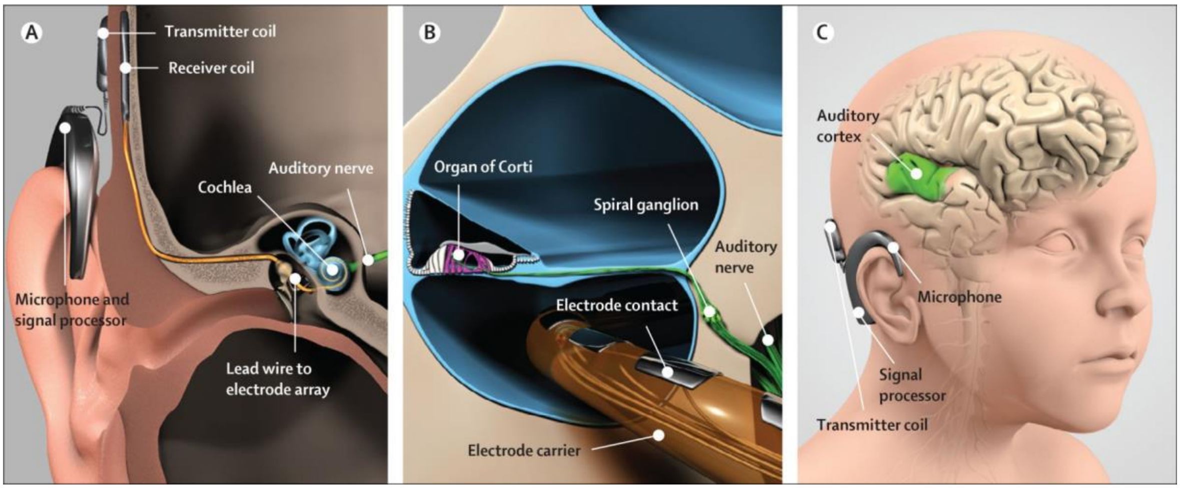


Figure 1.6 Components of cochlear implant. 1: microphone, linked to speech processor; 2: transmitter coil; 3: receiver electronics; 4: electrodes *array*; 5: acoustic nerve

The speech processor (with batteries) is located behind the pinna. Sound wave pressure variations are collected by microphone located above the pinna and converted into electrical signals variations by the speech processors; the electrical impulses are then led to the coil and transmitted through intact skin from the coil to the receiver electronics. The receiver-stimulator package (placed subcutaneously and fixed on the mastoid bone) receives electromagnetic signals and delivers them to an electrode array placed in the cochlea.

External coil and inner receiver are connected bidirectionally through a radiofrequency system. This connection is bidirectional and allows both the transmission of electrical stimuli and control of the interface functionality between the electrodes and the acoustic nerve.

The electrode array, placed in the tympanic scale, carries a variable number of electrodes according to type of implant and type of stimulation cycle. Stimulation can be configurated in a monopolar or a bipolar strategy. In the monopolar configuration each stimulating electrode uses remote electrode as a reference (usually placed outside the tympanic scale); in bipolar stimulation, the stimulating electrode has a nearby electrode as reference, providing more localization of stimulation areas (Cuda, 2009).

Modern CIs have up to 12-24 electrodes, can record evoked signals from the auditory nerve, and contain several speech-encoding algorithms that transform sound into electrical stimuli. Some algorithms allow signals to be directed to preselected auditory nerve regions.

Concerning electrodes' positioning, it can be extra-cochlear (around the round window) or intracochlear (on the tympanic ramp). Current devices widely use the intra-cochlear method which allows the positioning of the electrodes as close as possible to the nerve fibers, best preserving cochlear tonotopicity properties. In fact, the electrode contacts can exploit the tonotopic arrangement of nerve fibres, that is, the encoding frequencies system. In other words, different frequencies cause vibration in different points according the arrangement of nerve fibres, with low frequencies represented at the apex and high frequencies at the cochlear base. A functional cochlea acts as a real spectrum analyzer: each place of the cochlea vibrates at his own specific frequency leading a shift of hair cells. This so-called "place theory" was basic to develop modern multiple channel cochlear implants. The signal processor breaks down input signal into frequency components. The cochlear implant stimulation contacts are distributed longitudinally along the electrode array: this arrangement allows to exploit the tonotopic organization of the nerve fibers through the activation of different electrodes depending on the sound spectrum.

Activation of the implant generates an electrical response in selective auditory nerve fibres, which is carried to the auditory cortex and is finally interpreted as an auditory input by the brain. Once the neurons are stimulated, the input is conducted to the brain and interpreted as sound. As described in Chapter 3 of this work of thesis, cochlear implants, bypassing the damaged sensory organ, stimulate the neurobiological and neurocognitive substrates for speech and language processing, also promoting in infancy an integrated cognitive development.

Current CI technology provides physiologically useful intensity, frequency, and timing cues and a good perception of waveform (envelope) that are required for recognition of phonemes and speech comprehension. As a matter of fact, speech processing strategy is an extremely important aspect of CI systems and there are currently different signal processing techniques that can vary between different devices depending on the acoustic information that aim to more preserve (for example, the spectral characteristics, the waveform of the signal or the envelope) (for more informations on this topic, see for example

Choi & Lee, 2012). Nevertheless, current devices still cannot adequately represent the temporal fine structure of sounds: this point is not to underestimate, since it is linked to perception of F0, timbre and music appreciation (Wilson, 1997). Moreover, the encoding of very-low-frequency signals, such as the pitch of a voice, ca be limited (Kral, 2010). Thus, electrical stimulation induces a pattern of auditory nerve activity poorer than acoustic stimulation (Middlebrooks, Bierer & Snyder, 2005).

1.3.1 The role of binaural hearing and bimodal stimulation

When listening conditions are difficult (e.g. noisy environments), binaural hearing constitutes the primary listening condition to facilitate understanding of speech. Binaural hearing is fundamental for localization and orientation to sound source, also promoting environmental control and balance. As a matter of fact, the summing effect induced by two auditory input (from left and right ear) may provide an improvement loudness sensation in presence of noise up to 3 dB.

This phenomenon can determine the achievement of 6 dB at the supraliminal stimulation level (30-40 dB SL); moreover, the improvement in loudness sensation and in perceived volume is also accompanied by a greater sensitivity to differences in intensity and frequency, improving speech understanding both in silence and in noise.

Moreover, in binaural hearing condition the ear closer to the sound source can receive up to 20 dB louder input than the other ear, with an advantage for speech comprehension and an improvement in hearing sensitivity by ~3 to 10 dB. Although outer ear can provide some localization in monoaural condition by using spectral cues, it is less precise and poorer than with binaural hearing (Gordon, Henkin, & Kral, 2018).

Auditory perception environments and localization of sounds are based on central processing of Interaural Intensity Difference (IID); Interaural Time Difference (ITD) and Interaural Phase Difference (IPD).

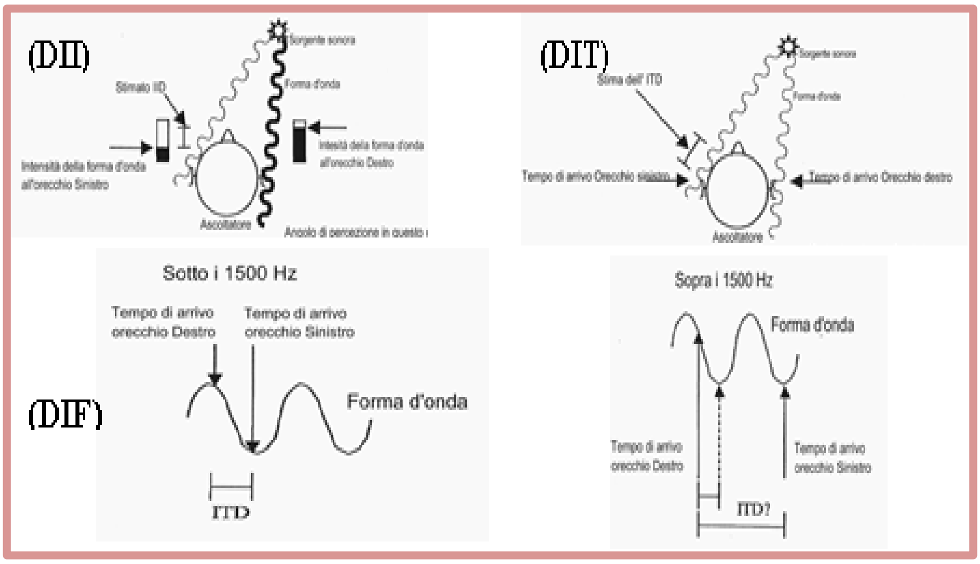


Fig. 1.7 Interaural wave sound differences: Intensity (IID), Time (IDT), Phase (IPD).

IPD refers to the difference in the phase of a wave that reaches each ear. In general, the more asymmetrical sound is in relation to patient's azimuth (0°), the easier localization, identification of sound and detection of differences will be. It depends on frequency of sound wave and on ITD. Once the brain has analyzed IPD, ITD and IID the location of the sound source can be determined with good accuracy. IPD, ITD, IID are induced by presence of the head, acting as a screen for sound ("shadow effect") and by natural distance of the ears from each other. IPD, ITD and IID are extremely useful not only in determining localization of sound, but also filtering the noise and catching up auditory target stimuli ("squelch effect"). Benefit of this suppression effect, a central phenomenon through which noise and its masking effect is reduced, allows a better perception of sound. This phenomenon of suppression depends on the ability of the central auditory system to spatially separate the source of speech and noise, to suppress the noisy sounds causing disturbance to the target stimulus to discriminate.

In contrast to binaurally, in monoaurally conditions, only one ear is compensated in the presence of significant hearing damage in both ears, causing an over time significant deterioration of vocal recognition capacity of the unprosthetic ear, both in adults and children. So, in case of monaural listening conditions, when sound comes contralaterally, because of the head shadow effect, the high-frequency units of sounds are attenuated, causing difficulty in hearing sounds in the deaf side (Kitterick et al., 2014). In this case, head constitutes a significant acoustic barrier with consequent difficulty following a conversation, understanding the verbal message and in localization and orientation to sound, difficulties in critical listening conditions, such as in noise environment, reduced ease of listening, greater auditory fatigue and greater cognitive effort, in some case leading to social disengagement and isolation (Tokita, Dunn & Hansen, 2014). Studies of bilateral cochlear implantation in children has shown that sound localization is improved by 18.5%; crucially, an average 20% improvement in the ability to hear speech against background noise has been reported under rigorous test conditions (Lovett et al., 2010).

It is now widely recognized that it is important not only to provide binaural amplification for bilateral hearing loss, but also to maintain the brain's ability to efficiently use inputs from each ear as early as possible. In fact, an early sensory restoration can retrieve the neural pathways useful for binaural hearing.

An early single-side deafness (SSD) can determine a risk of delayed language and learning skills (Bess & Tharpe, 1986; Bovo et al., 1988), because of listening effort especially in noisy and high reverberation environments (such as a classroom) with a significant increase of cognitive load (Rudner et al., 2018). In early development, functional magnetic resonance (fRM) studies have shown a modification of the neural networks normally activated in binaural children, in some cases causing the so-called “aural preference syndrome“ (Gordon, Henkin & Kral, 2015), with a more extensive representation of better ear on the auditory system and a worse cortical representation of the other ear.

In recent years, several studies have shown the benefit of bimodal stimulation, in both adults and children, and in quiet and noise (e.g., Ching et al., 2006; Devocht et al., 2017; Dincer D’Alessandro et al., 2015; Dorman & Gifford, 2017). Bimodal listening involves a combination of two different stimulation modes: electrical stimulation via a cochlear implant (CI) in one ear and acoustic stimulation via a conventional hearing aid (HA) in the contralateral ear. As a result of the broadening of CI indications to include patients who demonstrate considerable residual hearing in the contralateral ear and who benefit from conventional amplification in that ear, bimodal listening has become more frequently adopted at present (e.g., Ching et al. 2006, Sheffield et al. 2014; Devocht et al. 2017). Since existing, CI technology conveys degraded spectro-temporal acoustic cues in particular for the low frequency domain: the HA use combined with a CI has been shown to lead to improve speech perception especially in the presence of noise (e.g., Ching et al., 2006; Cullington and Zeng 2011; Dincer D’Alessandro et al., 2018a; Dorman & Gifford, 2017). Indeed, an increasing number of patients are adopting bimodal hearing including elderly patients.

As matter of fact, the current recommendation is that all CI users should wear an HA in the contralateral ear unless there is clear evidence to suggest that this will have a negative effect on auditory perception (e.g., Cullington and Zeng, 2011; Firszt et al., 2019; Dorman & Gifford, 2017).

1.3.2 Indications and contraindications for cochlear implants

Severe-profound hearing loss has wide-ranging implications for the affected person and often for patient's family and significant others. Evaluation for cochlear implant candidacy should be multifaceted, multidisciplinary, and embracing social and emotional needs, lifestyles, communication preferences, and expectations. The indications for the cochlear implantation gradually changed over the years, maintaining these overall guidelines:

- Cochlear implant outcomes cannot be totally predicted *a priori;*
- Selection of patients with high odds of improving their perceptive skills;
- Never have patients that perform more poorly with their cochlear implant than with hearing aids alone (Waltzman & Roland, 2006).

Anyway, at an international level, criteria for implantation and for access to CIs are not totally uniform, both for adults and for children. Most Countries of western world have national or local guidelines in place that govern candidacy rules for implantation (Vickers, De Raeve & Graham, 2016). It may depend on cultural and language aspect, and even on the model of service delivery and funding (Vickers, De Raeve & Graham, 2016).

For example, concerning the presence of obligatory guidelines or criteria, findings from an international survey across 17 different world regions (Vickers et al., 2016) highlighted that 10% do not have national or local CI guidelines and 20% have guidelines but decision for candidacy is down to the single clinical team.

Concerning presence of audiometric criteria, nearly 80% of countries base on audiometric guidelines both for children, whereas the percentage drops to 70% for adult implantation. Moreover, there is a variation in audiometric candidacy rules across countries. By way of example, in Belgium the average thresholds should be greater than 85 dB HL at 500, 1000, and 2000 Hz bilaterally; in Australia the average thresholds have to be greater than 70 dB HL >1500 Hz and the UK guidance indicates that thresholds should be greater than 90 dB HL at both 2 and 4 kHz bilaterally. The most accepted pattern of audiometric candidacy uses criteria in which the average thresholds should be greater than 75–80 dB HL at frequencies above 1000 Hz (Vickers et al., 2016). Concerning specific exclusion factors only 10–20% of countries have specific exclusion factors within their candidacy assessments, based on age, duration of deafness or etiology.

Italy is one of the countries that adopted candidacy and exclusion criteria for cochlear implantation, both for children and for adults, as published by the Italian Society of Otolaryngology (Quaranta, et al., 2009).

Indication for Cochlear Implant in Italy are as follow:

- *Adults (> 18 years):*

Adult patients, without age limits, with severe-profound hearing loss (average hearing threshold for the frequencies 500, 1000 and 2000 Hz> 75 dB HL) with speech discrimination of disyllabic words inferior or equal to 50% in free field in the best hearing aided conditions.

Material to administer disyllabic words, trisyllabic words and sentences (Quaranta et al., 1996) presented by voice of conversation or by voice recorded in auditory-only mode at the intensity of 70-75 dB.

A special category of CI candidates is represented by congenital or early onset of hearing impaired (HI) adults, who did not receive in infancy an early hearing aid rehabilitation therapy and an adequate and timely perceptual and verbal stimulation. In such cases, the development of the central auditory pathways and of the cortical areas assigned to verbal perception and production of language has been compromised: hence, in these cases, clinicians have to deserve extreme caution and an highly individualized approach to CI indication. Although these patients also do not represent an absolute contraindication to the cochlear implant, outcomes are often limited to detection of environmental noises, with a very poor increase in speech discrimination skills. An extreme attention beyond audiological variable is needed: the psychological and cognitive functioning, the level and quality of motivations and expectations, the presence of social and familiar support, a deep awareness of audiological and communicative limits due to a long auditory deprivation are all essential issues to take into account.

- *Children (aged up to 3 year):*

Age≥ 12 months.

Under 12 months of life, CI can be performed only in case of risk of early ossification of the cochlea (e.g. bacterial meningitis) or in those cases where the criteria of diagnostic certainty are met: severe-profound hearing loss (> 80-90 dB HL as average thresholds at 500, 1000 and 2000 Hz frequencies) ascertained with objective and behavioral methods (Table 1.3). Combination of all available methods and repeated measures for determining the threshold is required. Use of hearing aid in association with speech therapy rehabilitation for a period of not less than three to six months without evident perceptive and expressive benefits is an important clinical request.

- *Children (aged 3 -18 years)*

Severe-profound hearing loss (> 75 dB HL as the average for the 500, 1000 and 2000 Hz) ascertained with objective and behavioural methods.

Use of hearing aids in association with speech therapy rehabilitation for a period not shorter than three to six months without evident perceptive and expressive benefits.

Assessment of speech perceptions skills through administration of age appropriate tests.

Objective methods	Description
Otoacoustic emissions	A sensitive microphone is placed in ear canal for detection of mechanical energy propagated outward by metabolic activity in outer hair cells. OAE are an essential screening tool and it takes about 10 min per ear;
Auditory Brainstem Response	Measurement of electrophysiological responses to acoustic stimuli generated in auditory nerve and brain stem. ABR are used to determine hearing threshold; sedation is often required, and testing takes about 15 min for both ears; ear-specific results can be obtained
Tympanometry	Recording of middle-ear impedance as pressure in ear canal is raised or lowered. It is used to assess status of middle ear.
Acoustic reflex	Measurement of increased stiffness of middle ear due to contractions of middle-ear muscles in response to loud sounds. Useful for estimating hearing threshold or identifying sites of auditory dysfunction from middle ear to brain stem
Cortical evoked response	Measurement of physiological activity in auditory cortex. CERA is used to assess auditory functions, such as neurologic dysfunction and to monitor maturation of auditory system

Behavioural methods	Description
Observational audiometry	Changes in state of activity in response to sound in very young infants are assessed
Visual-reinforcement audiometry	Use of a head turn in response to an acoustic stimulus, which is then reinforced by a visual reward. VRA can be used in children since six months and gives frequency-specific and ear-specific information; should always be used as soon as possible to confirm objective tests

Table 1.3 Main objective and behavioural methods in children audiological assessment (adapted from Kral & O'Donoghue, 2010)

Absolute contraindications to cochlear implants are as follow:

- The absolute contraindications are represented by: cochlear nerve aplasia; unrealistic expectations and/or lack of motivation and patient's health conditions that contraindicate the execution of surgery under general anesthesia.
- Cochlear malformations in general are not an absolute contraindication for a cochlear implantation, while an intracochlear fibrosis, an auditory neuropathy or ossification may represent a relative or absolute contraindication.
- Other contraindications to CI are represented by chronic inflammatory pathology of the middle ear and some results of otological interventions. Vestibular nerve schwannoma is a relative contraindication. In fact, the application of a cochlear implant can only take place in selected cases of type II neurofibromatosis in which the residual neural functionality of the VIII cranial nerve has been documented as well as the anatomical integrity assessed by intraoperative electrophysiological methods (Ahsan et al 2003, Hoffman et al 1992).
- When deafness is associated to additional handicaps (such as visual, cognitive, mental, attention and learning deficits, autism, and pervasive developmental disorders) CI must be carefully evaluated. Nevertheless, these conditions are not absolute contraindication. Different studies have been conducted on these patient populations. The results document significant auditory benefit although progress is slow, unstable and depending on the severity of the concomitant deficit.

1.3.3 Personal considerations on understanding candidates' expectations and motivation: is it really important?

Among the absolute contraindications to cochlear implant, as indicated by Guidelines (Quaranta, et al., 2009), the cochlear nerve aplasia and a vulnerable general health precluding execution of general anesthesia are objective, easy to recognize and not-modifiable conditions. In this regard, it's necessary to point out that in selected case the CI surgery in local anesthesia is possible (Cuda, 2009).

The third contraindication underlined as absolute by Italian Guidelines (Quaranta, 2009) is the unrealistic expectations and/or lack of motivation. This contraindication is quite different from the others, it's less objective, not always easy to detect and in some case, it might be partially modifiable.

See, for instance, that the Handbook of Cochlear Implant (Cuda, 2009) in the section concerning the absolute contraindications to CI, report only the cochlear nerve aplasia, reclassifying the unrealistic expectations as moderate contraindication. The Guidelines for the application of the Cochlear Implant and the management of the Cochlear Implant Center (Quaranta et al., 2009) state that "the candidate for CI and his family must be informed of the technical characteristics of the CI, the risks associated with surgery and the presence of the CI, the limitations related to the CI and the potential benefits of applying the CI on perceptual and expressive skills" (p. 2; translation in English is mine).

Unfortunately, there is a major difference between being informed and deeply understanding; in addition, the competence required to analyse -in a non-superficial way- the patient's level of understanding, awareness, expectations and motivation would imply the presence of a professional capable of knowing how to read the patient from a psychological point of view. Unfortunately, however, although the Italian indications relating to the requirements of a cochlear implant center (Quaranta et al., 2009) provide for the presence or at least constant availability of a clinical psychologist, in clinical reality this indication often remains only at a theoretical level.

An evidence of the still current underestimation of psychological evaluation of cochlear implant patients is found in the paucity of papers written about it, compared to the number of reports on surgical and audiological aspects.

A PubMed search (June 21 2020) revealed 94 hits using the combination of the search terms "psychological assessment" and "cochlear implant". Replacing the term "psychological" with the terms "audiological" and "surgical" this number goes up to 9256 hits.

The assessment of psychological characteristics of candidates for cochlear implant (and of parents when candidate is a deaf child), together with an in-depth knowledge of their familiar, social, educational, environmental characteristics, should be common practice in all certified cochlear implant centers (Filipo, Bosco, Barchetta & Mancini, 1999).

Since 1993, formal and informal psychological assessment are an integral part of the Multidisciplinary Approach of Cochlear Implant Center of Policlinico Umberto I in Rome. The psychological assessment contributes to team decision about suitability of patients for implantation, also monitoring their progress and development across the years and through subsequent follow-up.

A psychological evaluation is thus required as part of, and complementary to, a global assessment of the child. It is aimed at defining a functional profile, including the neuropsychological and the affective development: the level and quality of skills, the relevance of these abilities and the relationship between functional skills and mental development, the dynamic organization of a personality in evolution. To get a picture of the child's developmental functioning, it is necessary to keep in mind both strengths and weaknesses, through a flexible approach oriented towards global assessment rather than pure psychometric testing. “Testing” refers to the application of a series of techniques, tests as objective as possible. The assessment sums up the entire evaluation process, where the use of tests is only a part. In this sense, the relationship becomes an essential element of this process. The relationship with the child and, necessarily, the relationship with that child's family. This aspect is always an essential tool of knowledge. It may result in a reasoned and complex process that first of all passes through the collection of information from various sources (parents, grandparents, teachers), the observation of child behavior and in interaction with caregiver and also the administration of valid and reliable tests.

In the preimplantation assessment phase, a crucial goal of psychological evaluation is to avoid the non-use of the implant. In the case of CI both in infancy and in adulthood, depending on patients’ biographical, audiological, medical, and psychological history, the pre-implant assessment protocol is structured and organized in distinctive and customized way. In order to prevent negative outcomes (e.g. non-use or underuse of CI) a careful psychological observation and evaluation is needed. Along with it, it is important to verify the presence of adequate cognitive, familiar, social, educational resources; to verify the existence of cognitive, space-temporal and behavioural prerequisites needed to accept and follow the rehabilitation protocol and to verify that expectations are realistic (Filipo et al., 1999).

In next chapters the issues related to psychological and neuropsychological aspects related to psychological care, evaluation, and rehabilitation of hearing impaired across the lifespan will be more in depth addressed.

Chapter 2. Hearing loss and cochlear implantation in infancy: the importance of a psychological scaffolding to the family

2.1 The role of family in intervention and care of deaf children

It sounds almost obvious that in paediatric cochlear implantation a clear and concrete involvement of parents in intervention and care process plays a crucial role: in case of congenital hearing loss in early childhood, an essential step after diagnosis should be directed to the reception, listening and taking care of the parents of the deaf child.

Nevertheless, how to make this assumption concrete is less clear and more demanding. In diagnostic and clinical path, leading parents to decide for a cochlear implantation, considering what is important to parents rather than to professionals, assumes crucial relevance in order to make informations really available and deeply understood.

From this point of view, psychological input offered by clinical psychologists in paediatric CI teams might be really useful. This means to approach in a biopsychosocial way to parents of a deaf child, taking into account all possible biological, psychological (thoughts, emotions, and behaviours), and social (e.g. peer relationships, family circumstances, and cultural issues) factors, as well as to understand their interactions.

The importance of putting families first in intervention and rehabilitation of hearing loss in infancy has been formally recognized in June 2012 when an International Conference took place in Bad Ischl (Austria) with the aim of coming to consensus on basic principles to adopt in family-centred early intervention with children who are deaf or hard of hearing (D/HH) (Moeller,Carr, Seaver, Stredler-Brown, & Holzinger, 2013).

The overarching concepts underlying the FCEI philosophy can be summarized as follows:

- Interventions based on explicit and validated principles and practices
- Flexible and holistic approach to family and D/HH child
- Recognition of strengths and skills of families
- Promotion of family wellbeing, joyful interactions and joyful parenting, engagement, self-efficacy and constant engagement in program.

At the end of the conference, a document with the foundational principles was produced, with the goal of promoting a widespread, international implementation of evidence-based principles for family-centred early intervention (FCEI) with D/HH children and their families (see Table 2.1)

Principle 1: Early, Timely, & Equitable Access to Services: Screening and confirmation that a child is D/HH will be effective to the degree that they are linked with immediate, timely, and equitable access to appropriate interventions.
Principle 2: Family/Provider Partnerships A goal of FCEI is the development of balanced partnerships between families and the professionals supporting them. Family–provider partnerships are characterized by reciprocity, mutual trust, respect, honesty, shared tasks, and open communication.
Principle 3: Informed Choice and Decision Making Professionals promote the process wherein families gain the necessary knowledge, information, and experiences to make fully informed decisions. This includes educating families regarding special education laws and their rights as defined by these laws. Decision making is seen as a fluid, ongoing process. Families may adapt or change decisions in response to the child's and families' changing abilities, needs, progress, and emotional well-being
Principle 4: Family Social & Emotional Support Families are connected to support systems so they can accrue the necessary knowledge and experiences that can enable them to function effectively on behalf of their D/HH children
Principle 5: Family Infant Interaction Families and providers work together to create optimal environments for language learning.
Principle 6: Use of Assistive Technologies and Supporting Means of Communication Providers must be skilled in the tools, assistive devices, and mechanisms necessary to optimally support the child´s language and communication development
Principle 7: Qualified Providers Providers are well trained and have specialized knowledge and skills related to working with children who are D/HH and their families. Providers possess the core competencies to support families in optimizing the child's development and child–family well-being
Principle 8: Collaborative Teamwork An optimal FCEI team focuses on the family and includes professionals with experience in promoting early development of children who are D/HH. Ongoing support is provided to families and children through transdisciplinary teamwork, whereby professionals with the requisite skills are matched to the needs of the child and family.

Principle 9: Progress Monitoring FCEI is guided by regular monitoring/assessment of child and family outcomes.
Principle 10: Program Monitoring FCEI programs evaluate provider adherence to best practices and include quality assurance monitors for all program elements

Table 2.1: Best Practise Principles in Family-centred interventions (adapted from Moeller et al., 2013)

As we can see, the first three principles focus on the importance of family involvement in decision making process: in the context of a Cochlear Implant Center, this means to offer families an adequate time and a psychological space to understand, ask for clarification, gain the necessary knowledge for making a decision about the better solution for their child. A child with hearing loss is not a life risk; to decide in favour or not of cochlear implant means also to take a decision concerning child life-long communication and educational options (Archbold et al., 2006).

This also mean, for professionals, to allow parents to acquire a quite clear state of mind to think and understand what is being explained, proposed, and offered to them. As a matter of fact, a state of mind clouded by pain, anger and guilt is not the optimal state for acting and choosing in full awareness. In clinical practice this can result in not complying with the third contraindication underlined as absolute by Italian Guidelines (Quaranta et al., 2009) - i.e. the unrealistic expectations and / or lack of motivation. Thus, we argue that it is essential to provide parents appropriate time, appropriate space and appropriate informations on which to base their decision to proceed with cochlear implantation for their child.

As we'll see in next paragraph, this also means to consider family within an integrated model in which "*brain science*" and "*mind scienc*e" could talk together in order to better understand and provide an efficient support to hearing impaired children and their parents.

2.1.1 A brain-mind approach to understand importance of early parent-child relations

From a neurobiological point of view, the brainstem - as modulating activation of Automatic Nervous System - can be thought as the physiological basis of the mind (Shore 2003). Phylogenetically and ontogenetically the brainstem is the oldest and earliest structure to emerge. Already active before birth and fully functional at birth, brainstem is poorly susceptible to experience and learning. Physiologically, the brainstem is a complex set of fibers and cells which, in part, function as a relay stations for transmission of informations from brain to spinal cord and cerebellum, and *viceversa*. It is also the region in which vital functions are regulated, such as breathing, cardiovascular functioning, digestion. In addition, the brainstem is connected to the activation of primitive reflexes, including those that provide the starting point for the process of infant attachment to her mother. Thus, for example, the first smiles of a new-born are reflexes controlled from the trunk, in order to attract those who can take care of him (Cozolino, 2002).

Cozolino (2002) points out that, unlike many organisms that are born with skills they use directly to survive, human babies survive is based on the abilities of their caretakers "to detect the needs and intentions of those around them" (p. 7). As a matter of fact, the secondary differentiation of human neuropsychological and cognitive functions depends largely on the early interactions and stimuli that the new-born receives, from a sensorial, auditory, olfactory and tactile point of view, such as contacts, caresses, mother's voice. In this sense, the brain can be seen as a social organ and evolution of the "social brain" as a method of survival. In fact, in a situation perceived as "safe", brainstem ventral vagus nerve restrains the activation of sympathetic nervous system: heartbeat slows down, breath becomes slower and deeper, muscles of head, neck are modulated, improving a "focused listening" and an effective social communication. On the contrary, in a condition perceived as "dangerous" , brainstem releases vagal brake, disinhibiting the SNS, preparing for a "fight or flight" response, speeding up the heartbeat and the breathing, increasing muscle tension, oxygenation, and vasoconstriction, tightening the jaw. In such situation, an effective social communication is impossible to achieve.

Thus, in this sense we can consider brainstem and ventrovagal circuit as the neuronal substrate of human "somatic self", that is, the original one's sense of self, deeply grounded in bodily experience. It allows us to be engaged in affective relations, to decode others' social and emotional messages from eye contact, facial expression, voice intonation, to respond properly and self-regulate physiological sensations.

At this point, assuming that brainstem represents the neuronal substrate of "somatic self", going on towards this short brain overview, undoubtedly the Limbic System can be thought as the neuronal substrate of human "emotional self". It must be said that the term "Limbic System" is just a descriptive term, used to lump a quite complex and multifunctional group of cerebral and diencephalic structure with interconnecting tracts (Figure 2.1). The cortical areas of the limbic system include: on the medial surface the *subcallosal gyrus and* *the cingulate gyrus; on the inferior surface there are the isthmus and the parahippocampal gyrus.* The major nuclei include the *septal nuclei, amygdala*, parts of the *hypothalamus*, parts of *thalamus* and parts of *reticular formation*.

Deeply on the medial surface of temporal lobe the *hippocampal formation* is placed, lying superior to the parahippocampal gyrus and extending from the caudal border of the amygdala to the caudal end of corpus callosum.

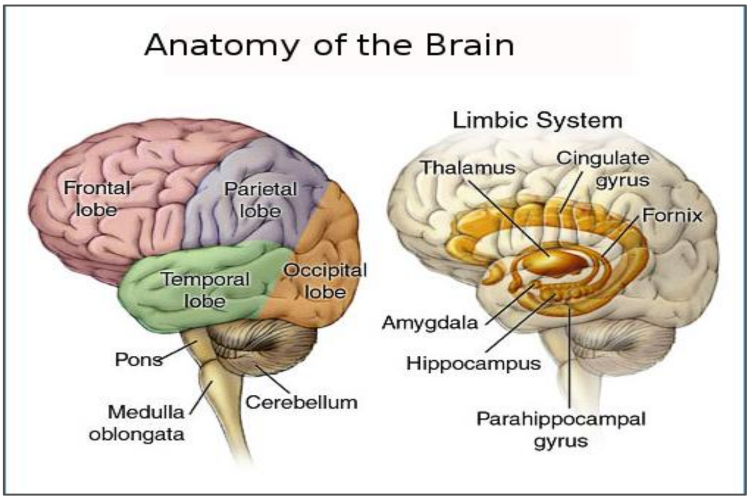


Figure 2.1: The Limbic System

The limbic system is where sensations and emotions are processed. It can be thought as the neuronal substrate of human “emotional self”.

Functionally, structures of the limbic system are implicated in mediating emotional expression and behavior and are also important for learning and memory functioning. In particular, the hippocampal formation is considered to be important for the transfer of recently acquired information (short-term memory) into a more enduring form (long-term memory). It receives input from the entorhinal cortex of the parahippocampal gyrus, the septal nuclei, and hypothalamus. The input from the entorhinal cortex "relays" information from the olfactory system, cingulate gyrus, orbital cortex, amygdala and temporal cortex.

The brain centers that mediate language and autobiographic memory (in particular the hippocampus) aren’t actually networked until 18-36 months: the impossibility of accessing memories of early childhood therefore seems to derive from a verbal inaccessibility to all the early affective and relational experiences that take place before having sufficient neural equipment to linguistically codify them.

At this regard, LeDoux, in his famous manuscript entitled “The Emotional Brain” (1996) underlined the parallelism of amygdala and hyppocampus system storing-information, storing different types of information on a specific experience.

Amygdala can be considered as the gateway to the limbic system and as a survival center (Rothschild, 2000) providing instinctive reactions to experience. In a fraction of a second amygdala is able to evaluate the sensory inputs, for example, the shape of a snake, the snarl of a dog, an angry face and, signaling to the brainstem to activate the sympathetic nervous system, translates the evaluation of the danger in a physical reaction (fight or flight). As LeDoux pointed out (1996), the amygdala records experience in the form of unconscious and prelinguistic emotional memories. These memories are like recorded traces that, outside of awareness and language, polarize our assessments of present experience (Wallin, 2007). In this sense, the amygdala has a low ability to differentiate, resulting in instantaneous and poorly discriminated reactions. This polarization is shaped and modulated via hippocampal formation that can be thought of as an “organizer of experiences in sequences and contexts”, enabling us to respond very differently depending on the context and the situation, calming down when the danger is recognized as a false alarm (Siegel, 1999). Studies on early traumatized patients offer evidence of a volume reduction in left hippocampus and a reduced development of the left hemisphere (Read et al., 2001; Read et al., 2014; Pitman et al., 2012). Furthermore, the volume of the corpus callosum is significantly smaller in traumatized subjects than in the control group. When a subject is asked to recall an emotionally disturbing experience, the hemispheric activity shows a clear shift to the right; on the contrary, if a subject is asked to recall a neutral experience, the shift is clearly to the left. A sort of "emotional seizure" seems to occur (Goleman, 1995) whereby the amygdala and its connections with the right hemisphere overwhelm the hippocampus and the ability to remember, encode and express with words the memories of a traumatic experience.

As mentioned above, the hippocampus begins to function from 18-30 months; previous experiences and learning imprinted in the amygdala are linguistically inaccessible, overgeneralized, and implicit memories. On the contrary, the memories that are imprinted with the help of the hippocampus, whose connections to the cortical centers continue to mature for many years, are contextualized, explicit and verbalized.

It is worthful to underline that many of these emotional memories take place in the preverbal phase, when the child's brain system is able to store only memories made of somatic sensations and emotions. Thoughts are associated only later, with the emergence the ability to think and express oneself through language (Young, Klosko & Weishaar, 2003).

In first years of life, brain system is able to store only memories made of somatic sensations and emotions. The impossibility of accessing memories of early childhood therefore seems to derive from a verbal inaccessibility to all the early affective and relational experiences that take place before having sufficient neural equipment to linguistically codify them.

One might ask: which are the main developmental consequences of what above mentioned?

- In the first years of life, constant and repeated non-attuned, poorly sensitive and unresponsive experiences can then inhibit the development of the hippocampus, leaving the amygdala's reactivity without adequate modulation. This might mean that, in some situations, emotional reactions can actually occur without involvement of superior brain systems. So for example when faced with the perception of a threat, the amygdala retrieves memory on an emotional level, triggering an anxiety or a fear reaction with a sudden bodily reaction.
- In this sense, a sensitive and responsive parenting may support and scaffold child to achieve the internal regulation necessary to support more complex social, emotional, and cognitive development (Landry, Smith, & Swank. 2007; Panksepp & Bieven, 2014). For example, in the first year of life, joint attention, i.e. the ability to share attention and interest towards external object or event , is strongly influenced by maternal scaffolding during early interactions (Legerstee, Fisher and Markova, 2005; Striano and Rochat, 1999). We know that between 9 and 12 months of life, infants develop a basic ability allowing child to use shared linguistic symbols: the ability to think to other as “intentional agents” and to share with him mental states. Anyway, during the second year of life, this “mind sharing” ability goes on through more and more mature communication skills, such as the use of the gesture of indication with requesting and declarative forms: it denotes not only the presence in the child of the concept of agency, but also the awareness of others’ different mental states that can be modified through interpersonal communication (Camaioni, 2003).
- It is therefore clear how the development of language begins before the possibility of using words and sentences and it rests significantly on the quality of social interactions that can support or not the early prelinguistic communicative intents. Oral language development necessarily requires early exposure to a language.

> Talking to a child is part of a communicative and relational framework made up of primarily emotional exchanges.

John Bowlby (1952;1958;1969), great relevant English psychologist and psychoanalyst, is considered the main founder of Attachment Theory, one of the century's most influential theories of personality development and social relationships. Bowlby originally based his enormous research on findings from neurobiology and ethology. Attachment is a key concept in developmental psychology, and it can be introduced as the deep and enduring emotional bond that connects one person to another across time and across space (Ainsworth, 1973; Bowlby, 1969). Bowlby (1958; 1969) primarily observed that the natural research of the child for proximity to the mother is deeply rooted in an innate protection need. “Attachment” can be defined as the everlasting emotional bond that characterizes the relationship between a child and an adult that protects, supports and comforts him, especially in situations that are perceived by the child as dangerous and stressful, such as fatigue, tiredness and disease (Cassibba & Van Ijzendoorn, 2005). According to Bowlby (1969; 1988) the tendency to develop an attachment bond towards one’s caregiver is innate and common to all primates. It is an emotional necessity, but it is also and primarily an innate need and it is phylogenetically determined: as a matter of fact, in any animal species, closeness to their mother simply increases their survival odds. In this sense, the behavioral attachment system assumes a central place in the evolutionary process, as well as nourishment and reproduction. Moreover, a child cannot choose whether or not to enter into a relationship with his parent: rather, it is an innate tendency, an innate need to attach himself to a primary figure that offers care, love, and protection.

In last century, a multitude of studies highlighted the influence of early attachment relationship on child's physiology, also determining level of sensitivity of “somatic self” to experience. By way of example, children that experienced relationships with sensitive and responsive mothers show a higher threshold of activation of physiological anxiety responses than children with unresponsive or unpredictable mothers (Lyons-Ruth & Jacobvitz, 1999).

The attachment bond is formed during the first 36 months of life: this is a highly sensitive period during which the need to feel loved, supported and protected is central to the child. As the child grows, other emotional needs are added and integrated with the nuclear need for protection: need for spontaneity and play, need for freedom of self-expression, need for autonomy, need for clear limits.

In other words, the growing child needs to perceive that the people who take care of him pay attention to him, admire him for what he is (and not only for what he does), are available to help him, give him trust, and know how to take control when necessary.

If the parent perceives himself as incapable, inadequate, or lacks sufficient psychological and /or social resources, the risk is that a reduced tuning, reduced sensitivity and low contingency become stable relation elements, highly interfering the development of a warm, welcoming, responsive and functional context for development of the child. Although the concept of *scaffolding* in psychology and pedagogy was born and widely used mainly about the educational context (Collins, Brown & Newmanm, 1995), over time its use has also extended to the area of dynamic and developmental psychology. In particular, studies from Attachment Theory (Ainsworth et al., 1978, Bowlby, 1958, 1969,1988) and Infant Research (Beebe et al., 2000; Beebe & Lachmann, 2003) highlighted the importance of providing an “emotional-affective scaffolding” to the child, primarily made up by shared experience of “mental contact” (Trevarthen, 1998), parental attunement and responsiveness. This type of "emotional scaffolding" is done through parent ability to empathically identify with the child's emotional state, with his needs and motivations, also adapting communication modalities, for example through rhythmic and prosodic variations (Stern, 1985; 1998).

A deafness diagnosis can deplete these parental responsive and adapting behaviours (Hintermair, 2006) because of complex interweaving of factors, such as the psychological status of the parents (Kurtzer-White & Luterman, 2003; Zaidman-Zait, 2008), the effect of limitations on the child’s access to sounds (Cole & Flexer, 2007), the way parents may modify their interactions with their children (Luterman & Kurtzer-White, 1999; Reichmuth, Embacher, Matulat, Am Zehnhoff-Dinnesen, & Glanemann, 2013).

Helping families of children with severe-profound hearing loss to address the psychological difficulties related to the diagnosis of deafness, supporting the relationship from the very first moments after diagnosis, could help to promote and strengthen a responsive care and prevent the establishment of dysfunctional caring and attachment modality.

Of course, there is no just one way to be a "good parent". Each parent builds a unique and irreplaceable relationship with his child according to several intra and interpersonal factors. Early studies on "parenting" going back to 50’-70’, aimed to outline and define an ideal good parental style, such as capable of promoting healthy development and good adaptation of children. Nowadays this model is largely outdated: there is not only a way to be a good parent. One of the main contributors to alternative ways to study parenting is Bornstein (1991) who underlined that parental ability is a multidimensional complex construct not reducible to parents' personal qualities. To paraphrase Aristotele we could state that "parenting is more than the sum of parents". A good parenting is therefore a complex construct that refers to the personal characteristics of the parent, his relational and social skills, his ability to respond to the needs of the child in order to guarantee his psychic, physical, emotional and social development, but also to the personal meaning attributed to be and become a parent, in turn conditioned by the personal experience and representation of himself as "being son".

Bornstein (1991) pointed out four aspects of parenting: the nurturant caregiving, which refers to the dimension of acceptance and understanding of the primary needs of the child (care and nurturing); material caregiving, which refers to the way in which parents organize and prepare the physical world in which the child develops; social caregiving, i.e. the parental ability to emotionally stimulate children by involving them in interpersonal activities; didactic caregiving, which includes the set of strategies that parents use to encourage child's comprehension of his environment.

Guttentag et al. (2006) identified four components related to a comprehensive and responsive parenting: the ability to respond to requests; the ability to maintain focused attention; the richness of language; the affective warmth.

A great number of studies highlighted that two qualities of maternal responses are highly predictive of an effective mother-child interaction: sensitivity and responsiveness, that in turn have their roots in two crucial parental skills, namely mind-mindedness and insightfulness. Mind-mindedness (Meins, 1997) can be defined as the adult's ability to attribute autonomous mental and intentional states to the child, while insightfulness is the ability to grasp what the emotional needs are at the basis of the child's behavior. To have these skills allows parent to be contingent, to offer warmth, sensitivity, protection and regulatory support to the child. On the other hand, an insecure attachment becomes a significant risk factor especially when other unfavourable conditions are associated (e.g. illness, disadvantaged socio-economic conditions, lack of social support).

The presence of a sensory disability - such as deafness - can crack parental self-confidence as "guide and secure base" (Bowlby, 1988), often leading out feelings of inadequacy, incompetence, frustration, rejection and guilt. The deaf child is a child who does not reflect the image of the "phantasmatic child", the child dreamed, expected, and imagined. Moreover, a child that can't hear the mother voice is at some extent as a "communication-rules breaker", breaching the familiar trans-generational communication model, threatening and compromising the natural responsiveness in parent-child interactions (Bosco, 2013).

2.1.2 Practical implications in the clinical approach to families of D/HH children

Theoretical and practical implications from these studies should deserve clinical attention in treatment and care of deafness in infancy, taking care first of all of parents' psychological status from the moment they receive diagnosis of deafness.

The first and early preverbal relationships give shape to the child's self: this means that intervening on the possibility that the deaf child can benefit from constant experiences of attunement, contingency and responsiveness represents a crucial objective in a rehabilitation approach that takes into account of the overall development of the deaf child.

All the above shows the importance to offer a psychological scaffolding to parents even before the proper prosthetic solution; the importance to provide parents a proper time and a proper psychological space. It means to consider intrapersonal and interpersonal dynamics activated when parents receive diagnosis of deafness for their child. At the same time, it means to wonder what the consequences on their responsiveness and ability to scaffold child experience might be. Moreover, it is crucial to consider how and to what extent the alteration in natural responsive behaviours and psychological status may deplete not only linguistic but even emotional development.

In a way, it is necessary to offer parents what they shall provide to their child: a safe haven to recognize, accept, express and regulate fear, sadness, anger and any other difficult emotion; and, together, a safe guide that can gradually lead them to explore and get to know their child, to recognize his needs, his unique characteristics, his strengths and also his weaknesses.

Not providing properly time and space to parents may lead to an "interactive breakdown". It means risking that the early parents-child interactions remain a still silent and not tuned desert, with likely negative effects on psychological, cognitive and linguistic development.

The optimal timing for offering this psychological scaffolding to parents should preferably be the period immediately following the diagnosis of hearing loss.

Some key elements of the initial psychological scaffolding to family, as offered by Cochlear Implant Center of University La Sapienza are given below:

- *A space to cry.* Reception of the family after being diagnosed with deafness of the child. The psychologist welcomes parents in an atmosphere of total acceptance and respect for any personal emotional reaction, immediately trying to lay the foundations for parents to feel they are in a "safe place", where any emotion is accepted and is welcome, especially emotions not recognized, not recognizable or perceived as unacceptable as source of feelings of guilt.
- *A frame of meaning.* Helping parents to accept and contain painful emotional experiences and promote the creation of a frame of meaning, sharing and expressing emotions, doubts, thoughts, anxiety and fears.
- *Scaffolding communication.* Supporting and promoting communication between parents and child: helping the parent to recognize and understand the importance of the continuity of communication with their child, offering clarifications, providing examples, helping parents to perceive the child's signals and respond appropriately. In some case the analysis of videotaped frames can be useful in order to return to parents an image of themselves as capable and competent, and to reinforce in them the predisposition to see the good and the resources of their child, beyond the disability.
- *Scaffolding emotions*. To help parents to respond to the emotional requests of the child: focus on profound meaning of the parent-child interactions, focusing on child attempts to communicate, improving parenting sensitivity and responsiveness through work on specific skills, such as observing the child, recognizing his or her needs, responding in a contingent way, reflecting on personal psychological dynamics, supporting attunement, resonance, and empathy.

The caregiver has a fundamental role in encouraging the infant's involvement in the communicative exchange, empathically identifying himself with his moods and motivations, and offering him adapted communication methods with rhythmic and prosodic variations (Stern, 1985). Fundamental rhythm of the repeating movement, short expressive explosions, repetitions of groups of rhythmic movements, modulation of the intensity of the expression also have the role of "amplification" of the emotions and experience lived by the infant. Rhythmic and prosodic variations constitute privileged channels for the transmission of emotions, and it is precisely the passage of emotional expressions from mother to new-born and from new-born to mother that defines a "close mental contact" between partners (Trevarthen, 1993). On the contrary, the emotional disturbances of one of the two partners, for example maternal depression, can block the possibility of success of the intersubjective experience, with possible negative consequences on the psychological growth of the child (Tronick, 1989).

To conclude this brief presentation of CIC approach, as Kral and O'Donoghue (2010) has sharply underlined, we should always think to deafness as a family matter. “Listening to parents’ views and valuing their roles and input remain the most helpful clinical intervention; thus, time for parental engagement should always be given priority” (p. 1442).

2.2 Parent training and communication empowerment of children with cochlear implants

Abstract

Deaf children with cochlear implants (CIs) need a supportive family environment to facilitate language development. The present study was designed to assess the effects of parent training (PT) on enhancing children’s communication development. The PT was based on the “It Takes Two to Talk” model, with specific adaptations for families of deaf children. Before and after the PT, 14 participating families and matched no-treatment controls were assessed using the Parent Stress Index and Cole’s interaction checklist. The children’s language was assessed with the MacArthur–Bates Communicative Development Inventory and, after 3 years, with the Boston Naming, the Peabody, and the Test for Reception of Grammar–Version 2 (TROG-2). The families’ quality of interaction and the children’s language increased significantly more in the trained group than in controls and differences were still present after 3 years. The parents seemed to benefit from PT that focused on strategies to empower and promote communication skills in children with CIs.

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Introduction

In the last decade, the role of parents in the habilitation process with their children who are deaf or hard of hearing (D/HH children) has been emphasized. Family-centered programs have been designed to increase families' active participation (Hintermair, 2006; Moeller, Carr, Seaver, Stredler-Brown, & Holzinger, 2013). Specialists strongly recommend family-centered programs due to the growing amount of research that emphasizes the influence of family variables such as parental involvement and communication style in determining long-term outcomes after a deafness diagnosis and cochlear implantation (Cole & Flexer, 2007; Moeller et al., 2013). Indeed, parents play an important role in the communicative development of their children. During daily interactions, parents may naturally offer wide and varied child-directed words and syntactic structures (Cole & Flexer, 2007; Landry, Smith, & Swank, 2006; Quittner et al., 2013; Sarant, Holt, Dowell, Rickards, & Blamey, 2009) that contribute to children's acquisition of new words and morphosyntactic elements (e.g., Hadley, Rispoli, Fitzgerald, & Bahnsen, 2011).

However, a high rate of exposure to linguistic input alone is not sufficient to support communicative development. A key element of communicative learning is parental responsiveness, or the ability of parents to "tune in" to their children, and to recognize their child's communicative attempts and provide contingent responses (Suskind, Suskind, & Lewinter-Suskind, 2015).

Parental responsiveness supports a relaxed interpersonal climate in which children may be encouraged to communicate with the adult in a "back and forth" manner that is optimal for learning (Nittrouer, 2010). Parents who are verbally responsive support advances in children's language when they provide labels and comments for objects and events under joint attention and when they imitate and expand children's production of sounds and words (Tamis-LeMonda et al., 2001). In this way, children are likely to match linguistic symbols to their referents and reinforce the social-communicative functions of language (Tamis-LeMonda, Kuchirko, & Song, 2014). Children with more responsive parents, when compared with less responsive ones, may reach the vocabulary burst milestone earlier and have broader expressive and receptive vocabularies (Bornstein, Tamis-LeMonda, Hahn, & Haynes, 2008; Hart & Risley, 1995; Masur, Flynn, & Eichorst, 2005; McGillion et al., 2013;

Tamis-LeMonda et al., 2001). Moreover, higher levels of parent responsiveness seem to facilitate greater growth in social, emotional, and cognitive competence (Landry, Smith, & Swank, 2006; Merz et al., 2015; Walker, Greenwood, Hart, & Carta, 1994).

Parental communication is also essential for language development in DHH children. Research has associated different types of communicative behaviors played out by parents with better linguistic outcomes in their children. Studies demonstrate that listening strategies (e.g. reduction in environmental noise, speaking within earshot, the use of acoustic highlighting, or auditory hooks) create listening environments with favorable speech-noise ratio and facilitate auditory attention and processing (Estabrooks, MacIver-Lux, & Rhoades, 2016). The use of interactive strategies (e.g. following the child's lead and sharing joint attention) by parents after a deafness diagnosis was positively associated to spoken language skills of their children at three years of age (Cejas, Barker, Quittner, & Niparko, 2014). Responsive strategies (e.g. warm and contingent responding, balancing conversational turns, positive regard and respect of child's autonomy) together with facilitative language techniques (e.g. modeling child productions, commenting, expanding, recasting, adults' use of parallel talking) have been identified as predictors of the development of expressive and receptive language when both are used during play (Cruz, Quittner, Marker, DesJardin, & CDaCI Investigative Team, 2013; Quittner et al., 2013) and joint book reading (DesJardin et al., 2014).

A deafness diagnosis can alter most of these natural responsive behaviors (Hintermair, 2006) because of three key elements: the effect of limitations on the child's access to sounds (Cole & Flexer, 2007), the way parents may modify their interactions with their DHH children (Luterman & Kurtzer-White, 1999; Reichmuth, Embacher, Matulat, Am Zehnhoff-Dinnesen, & Glanemann, 2013), and the psychological status of the parents after a diagnosis of deafness (Kurtzer-White & Luterman, 2003; Zaidman-Zait, 2008).

With respect to the first aspect listed above, most DHH children who use cochlear implant(s) (CI) or hearing aid(s) (HA) typically show speech perception close to 100% (Geers, Nicholas, & Sedey, 2003; van Wieringen & Wouters, 2015). However, the limits of technology involve a difficult perception of speech in conditions such as: the presence of background noise (Yang, Hsieh, & Wu, 2012), an increased distance from the sound source (Whitmal & Poissant, 2009), an intensity of the primary signal below device threshold (Davidson, Geers, & Nicholas, 2014) and when the number of communication partners involved in an interaction increases (Tobey, Shin, Sundarrajan, & Geers, 2011). These factors reduce the quantity of words and sentences the child can hear and understand (van Wieringen

& Wouters, 2015; Yang, Hsieh, & Wu, 2012) and may negatively affect the opportunity to "overhear" speech prevent consistent access to auditory linguistic information. It is therefore crucial for parents to learn how to support their children's listening environment (Cole & Flexer, 2007).

The second aspect concerns the modifications of communication modalities between hearing parents and their DHH children. A diagnosis of deafness can reduce parents' self-confidence in child education and child-oriented behavior (Luterman & Kurtzer-White, 1999; Reichmuth et al., 2013). DHH children's limited responses to oral language may negatively influence parents' communication initiatives and generate frustration and confusion (Harrigan & Nikolopoulos, 2002; Spencer & Meadow-Orlans, 1996). Consequently, the establishment of joint attention and tuned communication may be less successful (Nowakowski, Tasker, & Schmidt, 2009) and parents may become more intrusive, directive, and less flexible during interactions (Aragon & Yoshinaga-Itano, 2012; Cole & Flexer, 2007). This can reduce the number of conversational exchanges and parents' responsiveness to their child's communicative attempts (Aragon & Yoshinaga-Itano, 2012; Nittrouer, 2010; VanDam, Ambrose, & Moeller, 2012).

Finally, as to third aspect, feelings of sadness, grief, and anxiety may characterize the psychological status of parents after a diagnosis of deafness. Discrepancies between parents' expectations and their child's actual competencies, everyday communicative failures, educational concerns, and the need for knowledge regarding how to better promote language may reinforce these feelings (Kurtzer-White & Luterman, 2003; Zaidman-Zait, 2008). For example, some research has found that the level of parental distress is inversely associated with DHH children's communication and social and emotional development (Quittner et al., 2013). In the absence of adequate resources, a higher initial level of parental stress was predictive of stress maintenance as the child grows (Lederberg & Golbach, 2002) and accounts for up to 30% of the variance in receptive language development (Quittner et al., 2010).

To reduce the negative impact of these three processes and to help parents rediscover their self-confidence, parental involvement has become constant within individual sessions during Auditory Verbal Therapy (AVT) (Estabrooks, MacIver-Lux, & Rhoades, 2016) and Natural Aural Oral Education (Clark, 2006), and it was the core of structured group sessions in specific Parent Training (PT) programs (Glanemann, Reichmuth, Matulat, & Zehnhoff-Dinnesen, 2013; Harrigan & Nikolopoulos, 2002; Reichmuth et al., 2013).

In particular, the first experiences in group PT for families of DHH children with hearing aids or CIs were led by Harrigan and Nikolopoulus (2002) and Glanemann et al. (2013) and were based on the It Takes Two to Talk (ITTT), a program developed for children with language delay by the Hanen Center in Toronto, Canada, with the aim of empowering family-child interactions and making parents the language facilitators for their children (Manolson, 1992; Pepper & Weitzman, 2004). Researchers adapted ITTT's principles to the special needs of families of DHH children (Glanemann et al., 2013; Harrigan & Nikolopoulos, 2002) and used them to enhance parental responsiveness to the communicative signals of their child. In order to achieve this goal, parents learned to apply specific strategies such as observing, waiting, and listening to the child's communication attempts before responding appropriately, mirroring vocal and non-vocal signals, such as movements and actions in order to create joint attention and balanced turn taking. The researchers also placed an emphasis on the adaptation of spoken language during interactions according to the child's developmental stage of communication, while adding information that could help the child increase their linguistic skills.

After the training, Harrigan and Nikolopoulos (2002) observed that parents' communicative behaviors shifted toward a more responsive method of interaction with their children. Their study did not include a control group of untrained parents and did not analyze the effect of such changes on the development of DHH children's communicative skills. Further, after a 12 month interval, authors observed that parents demonstrated a reemergence of excessive initiating behaviors which suggested that the parents' tendency to control interactions was subject to relapse and that the initial positive effects of PT could diminish if they received no further appropriate guidance.

Glanemann et al. (2013) developed the Muenster Parental Programme (MPP) which was inspired by ITTT and integrated with the Natural Auditory Oral Approach for DHH children (Clark, 2006) and with Play and Learning Strategies (Landry, Smith, & Swank, 2006). The program was used in a group of families with DHH children who had varying degrees of deafness. The authors studied changes in parents' communication style and the consequent effects on the development of their children's vocalization. This study included a control group of untrained parents and their DHH children. They confirmed that group PT was effective as a method to increase communication-enhancing behaviors and to reduce communication-inhibiting behaviors in PT parents. By the end of the training, the researchers found that the trained parents were significantly more attentive and responsive to the child's vocal signals than untrained parents. Children of parents in the PT group demonstrated significantly more vocalizations when compared to the children in the control group.

Suskind et al. (2016) conducted a further study that utilized the ASPIRE curriculum to improve the listening and linguistic environments of DHH children from families with low socio-economic status. Their results showed a positive impact of the program on parental knowledge concerning children’s language development and on the quality of the parents’ linguistic exchanges with their children. The study detected no positive effects on the DHH children’s language. Further, after 3 months from the end of PT, the improvements in parental quality of communication had disappeared from their interactions.

Given the paucity of studies on such an important topic, the present study was implemented with the purpose to shed further light on the efficacy of an ITTT model-based group PT program (Manolson, 1992; Pepper & Weitzman, 2004) in order to enhance the quality of interaction between a group of hearing parents and their DHH children with CIs. It was designed as a prospective clinical study, with PT and control groups matched for sociodemographic and audiological characteristics of the parents and children so that we could evaluate how changes induced by group PT impacted the parent’s level of stress and the development of children’s communications skills. We expected that PT could enhance parents’ responsive behavior, optimize their communicative performance in daily interactions, and contribute to their children’s listening and language growth. We also expected that more responsive and relaxed interactions would reduce stress, negative coping behaviors, and feelings of burden, which would improve the parents’ self-confidence. Finally, we anticipated a significant improvement in the rate of language acquisition in children whose parents received PT compared to children whose parents did not.

Method

Study design

The study was implemented as a prospective clinical study in which participants in the study and the control groups were matched for parental socio-economic status (SES) and education level, children’s chronological age (±2 months), hearing age (±2 months), pre-implant PTA at 250-4000 Hz, and language level. The absence of other associated disabilities in the children was verified through the General Developmental Quotient measured by the Griffith Scales of Child Development (Huntley, 1996; Luiz et al., 2006); the minimum acceptable value was set at 80, defined by the scales as “below average” (Huntley, 1996; Luiz et al., 2006). The parents who belonged to the study group attended the parent training program described above, while the control group did not. In both groups, the children attended aural-oral habilitation therapy. Both groups were assessed with the same timing: both parents and children during the immediate pre-training phase (within one month of the beginning of parent training) and at the end of the training (within one month of the end of parent training); children received a further control assessment at the three-year follow-up.

Participants

Tables 1 and 2 show the demographic and baseline characteristics of the parents and children, respectively. The study group included 22 parents of 14 children who were profoundly deaf and received cochlear implant(s) (CI-children) at the Cochlear Implant Center of xxxxxxxxx. It included 14 mothers and 8 fathers, with an average chronological age of 35.4 years (*SD* 5.4) and 40.4 years (*SD* 4.3), respectively. In 8 families, both parents chose to participate, and for the remaining 6, only the mothers attended the parent training as the fathers found it difficult to obtain permission from their employers. The parents completed a short questionnaire about their income and education level, defined as the number of years in formal education. The income bracket information was used to define the family's socioeconomic status and were determined according to the Italian National Institute of Statistics -ISTAT (https://www.istat.it/it/files/2017/12/Report-Reddito-e-Condizioni-di-vita-Anno-2016.pdf). Two families had an upper-middle SES, 7 had a middle SES, 2 had a lower-middle SES, and 3 had a low SES. The education level of the parents was on average 15.5 years of study (*SD* 3.2). Four parents had only 8 years of study. At the time of the study, the children of the trained parents (seven females, seven males) had a chronological age of 25.6 months (*SD* 6.3) and a hearing age of 7.8 months (*SD* 5.1).

The control group included 22 parents, 14 mothers and 8 fathers, with an average age of 37.8 years (*SD* 5.3) and 44.5 (*SD* 4.9), respectively, and their 14 CI-children (seven females, seven males) with a chronological age of 26.2 months (*SD* 6.2) and a hearing age of 8.1 months (*SD* 4.3). The SES of the parents in the control group was similar to that of the study group: 2 families had an upper-middle SES, 8 had a middle SES, 2 had a lower-middle SES, and 2 had a low SES. The parents had, on average, 13.7 years education (SD 3.8). Four but 4 of them had only 8 years of study.

Families were recruited when accessing the CI center for the routine follow-up appointment. The selection followed the order of arrival at the center and ended when all well-matched subjects were identified.

Treatment

The parent training (PT) was based on the ITTT model (Manolson, 1992; Pepper &Weitzman, 2004), with additional information on deafness, hearing devices, hearing environment, and strategies to improve listening skills that was relevant to families of DHH children. This further information was adapted from AVT (Estabrooks, MacIver-Lux, & Rhoades, 2016), the Natural Aural-Oral Approach (NAOA) (Clark, 2006), and Learn To Talk Around The Clock (Rossi, 2003). Two of the authors held ITTT certifications, and one was in the process of AVT certification. The PT program included both a primary (Phase 1) and a maintenance phase (Phase 2). Table 3 illustrates the detailed structure.

During Phase 1, nine group training sessions in the clinic and three individual sessions at home were performed, as indicated by the ITTT (Manolson, 1992; Pepper &Weitzman, 2004). A training group could include a minimum of three to a maximum of five families. Both parents were invited to participate. Each participating family received an illustrated handbook, observation sheets, and checklists to support the learning process. Group sessions took place every 15 days for the duration of the program. Each group session lasted 2.5 hours and focused on a specific theme. A ninth group session was planned to sum the PT's experience. The primary phase lasted a total of 4.5 months.

Various teaching strategies for the primary phase included problem solving activities, brainstorming, role playing, and video analysis, according to the instructions in the ITTT manual (Manolson, 1992; Pepper &Weitzman, 2004). Parents attended individual sessions between group sessions, which allowed them to practice the new strategies with their child based on previously agreed communication objectives. Video modeling (Pepper &Weitzman, 2004) was used to help parents transfer the knowledge they learned during group sessions to their everyday interactions with their child.

The observation was guided by the sheets and checklists provided by the ITTT program. The specialist provided the parents with feedback to help them apply the less-used strategies (e.g. waiting more, expand child's language, etc.) and played back video recordings during subsequent group sessions to create a dialogue between the parents and to allow all the families to learn from one another's experiences.

Table 4 illustrates the themes and the strategies presented in each group session. The first five group sessions aimed to support the parents' acquisition of new skills to improve listening environments and listening skills, to appropriately manage hearing devices, to better understand communication and language development, to use effective strategies to facilitate communication and language, and to apply these strategies in everyday activities and routines. In the last three sessions, parents learned how to apply the presented facilitation strategies to help their children achieve specific and realistic communication goals in the context of play, early literacy, and music.

At the end of Phase 1, an individual reinforcement and maintenance phase (Phase 2) was implemented to avoid the parents' regression to previous communicative methods or unbalanced interactions. The reinforcement and maintenance phases for all the families included an individual session per month for 6 months after the end of the primary phase. These individual sessions followed the same structure as the three individual home visits performed during the PT program.

Parent assessment

Parents were assessed with the Parent Stress Index-Short Form (PSI-SF) (Abidin, 2003) to evaluate the level of stress, and the Communication-Promoting Behaviors Checklist for Caregivers, to evaluate the quality of their interactions with their children (Cole, 1992).

The PSI-SF is a parental self-report inventory designed as a screening and diagnostic instrument for the early evaluation of stress in the parent-child relationship and the identification of dysfunctional behaviors during parent-child interactions that may interfere with normal infant development and functioning (Abidin, 2003). The PSI-SF is a 36-item version derived from the full-length Parenting Stress Index inventory (consisting of 120 items). Authors developed the PSI-SF based on the assumption that some degree of parenting stress is to be expected and that the total stress perceived by parents might vary due to child and parent characteristics and in relation to contextual characteristics, such as lack of social or familial support or how difficult it is for the family to access the necessary services. Parents answered all 36 items on the PSI-SF using a 5-point Likert scale (responses ranged from Total Disagreement to Total Agreement) and items used the same wording as the PSI Long Form. This questionnaire is easy to administer, does not take much time, and provides a Total Stress (TS) score that indicates the overall level of stress a person feels in their role as a parent based on three subscales: Parental Distress (PD), Parent-Child Dysfunctional Interaction (P-CDI), and Difficult Child (DC). The PD Subscale consists of 12 items and yields a measure of perceived parental distress in relation to personal factors not strictly related to the parent-child relationship, such as discord between the parents, feelings of incompetence, poor self-confidence, or lack of social support. The P-CDI Subscale consists of 12 items that assess the level of satisfaction of parents in their interactions with the child, and also consider the parents’perceptions of non-compliance, unsatisfactory or displeasing interactions, negativity, and rejection. The DC Subscale consists of 12 items that measure the parent’s perception of their child’s temperament, behavior, and compliance, and the extent to which the parent perceives the child as easy or difficult to take care of. Authors consider PSI-SF scores typical if they fall between the 15th and 80th percentile and consider stress scores clinically relevant when they are above 85 for the P-CDI subscale and above 90 for the other subscales. The PSI-SF also includes a Defensive Responding (DR) scale that is not included in the Total Stress raw score. Low scores on this scale (below 10) might indicate defensive answers that minimize problems so that the parent appears competent, or that the parents have poor investment in or consciousness of stress in their parenting role.

The Communication-Promoting Behaviors Checklist for Caregivers evaluates the quality of interaction based on a video-recorded sample analysis (Cole, 1992). This checklist provides a framework that is useful when we consider the appropriateness of the parents’ role in communicative interactions with their young DHH children and outlines the major components of optimal communication-promoting caregiver behaviors (see Appendix for the full checklist). It includes 22 items: the first 5 items (Sensitivity to Child - SENS) refer to the caregiver’s awareness of the child’s way of being and to their ability to adapt to the child in a supportive manner; items 6 to 11 assess the caregiver’s conversational behaviors when they respond to the child (Response - RESP); items 12 to 15 outline the conversational behaviors in terms of how well the caregiver is able to establish and maintain shared attention with the child (Shared Attention - SA); items 16 to 22 refer to general conversational behaviors (General - GEN). A numerical rating from 1 (behavior rarely observed) to 7 (behavior often observed) is given to each item based on a videotaped sample of the parent-child interaction.

Two trained speech therapists rated the interactions of all parents in a blind setup, where they were not aware which parents participated in the parent training group. The raters assessed each item, and then calculated a mean score for all 22 items (Overall Score-OS) and for each sub-category of the checklist (SENS, RESP, SA, GEN).

Child assessment

A speech therapist assessed the children’s linguistic skills pre-and immediately post-PT based on the MacArthur-Bates Communicative Development Inventory- MCDI (Fenson et al., 1993 - Italian edition, Caselli & Casadio, 2007) Gestures and Words Form, a widely recognized parent reporting tool to assess children's early language skills development for clinical and research purposes. The Gestures and Words Form is available for children ranging from 8 to 17 months of chronological age and was selected according to the average hearing age of the study and control samples (7.8 months (*SD* 5.1) and 8.14 months (*SD* 4.3), respectively.

Language abilities three years post-PT were assessed using the Italian versions of three standardized language tests: the Boston Naming Test (BNT) (Kaplan, Goodglass, & Weintraub, 2000; Riva, Nichelli, & Devoti, 2000) to assess lexical production; the Peabody Picture Vocabulary Test (PPVT) for lexical comprehension (Dunn & Dunn, 1981; Stella, Pizzoli, & Tressoldi, 2000), and the Test for Reception of Grammar (TROG-2) (Bishop, 2003; Suraniti, Ferri, & Neri, 2009) for morpho-syntactic comprehension. The BNT originally assessed adults' lexical skills in production and was later standardized for children (Kindlon & Garrison, 1984; Guilford & Nawojczyk, 1988; Riva, Nichelli, & Devoti, 2000) as researchers considered it an useful tool to assess children with learning and language disorders (Wolf & Obregon, 1992). The TROG-2 (Bishop, 2003; Suraniti, Ferri, & Neri, 2009) is a fully revised and newly standardized version of the widely used TROG, which was originally developed to investigate morpho-syntactic comprehension skills in children. The raw scores were used for a direct comparison between the PT and control groups. This was possible because the two groups were properly matched and the differences in chronological age and in the level of their basic language were not statistically significant. The use of raw scores allowed the recording of children's use of new words and which morpho-syntactic structures they understood and produced in the same intervals and offered the possibility to perform a quantitative analysis of the differences between the two matched groups.

Speech recognition was assessed in quiet and noisy (speech noise) environments with standard, phonetically balanced Italian disyllabic words and sentences for pediatric populations (Cutugno, Prosser, & Turrini, 2000). A practice list preceded each 10-item list. The items were administered in a sound-proof room via a loudspeaker that was placed 1-m from a table where the child sat next to a speech therapist. Stimuli were presented at 0° azimuth, with speech at 65 dB and noise at 55 dB (Speech/Noise ratio +10). The score was calculated as the percentage of correctly repeated words.

Results

Families

All the included subjects completed the study procedures; there was neither drop-out nor loss to follow-up. There was no significant difference between the PT and control groups in the demographic characteristics of either the mothers or fathers (chronological age, education level) (Table 3).

While the psychological characteristics and stress levels of the mothers did not differ between the PT and control groups, fathers belonging to the PT group had lower values for the Cole's Communication-Promoting Behaviors Checklist for Caregivers overall score (OS) and the Sensitivity (SENS), Conversation in Response (RESP), Shared Attention (SA) and General Conversation (GEN) sub-scores (p-values<0.05). However, there was no difference in the fathers' stress levels between the PT and control groups (see Table 3).

At the end of the parent training period, we found an overall improvement in the OS, the SENS, RESP, and SA scores, and the overall GEN regardless of the group, as revealed by a significant time effect, in both the mothers (F1,26 ranging from 146.0 to 336.4; p<0.001) and fathers (F1,14 ranging from 13.6 to 22.2; p<0.01). However, the PT group achieved better outcomes than the control group, with significant time-by-group interaction effects for all measures in both the mothers (F1,26 ranging from 74.5 to 160.1; p<0.001) and the fathers (F1,14 ranging from 11.8 to 5.1; p<0.05). Effects sizes exceeded 0.8; probability ranging from 84% to 96% (see Table 5).

In contrast, the stress levels did not change over time, as revealed by a nonsignificant time effect for both the mothers (F1,26 ranging from 0.1 to 1.3; p>0.3) and the fathers (F1,14 ranging from 0.4 to 2.4; p>0.2). The intervention did not affect the stress level, as revealed by a nonsignificant time-by-group interaction effect for both the mothers (F1,26 ranging from 0.1 to 3.7; p>0.05) and the fathers (F1,14 ranging from 0.1 to 3.6; p>0.05).

Children

There was no significant difference between the PT and control groups in demographic and clinical characteristics (see Table 2).

In both PT and controls, the MCDI showed an overall improvement in all the measures over time. In fact, in all children there was an increment with a significant time effect ($F_{1,26}$ ranging from 79.7 to 208.8, p-values<0.001). However, the PT group achieved better outcomes than the control group, with significant time-by-group interaction effects for all measures ($F_{1,26}$ ranging from 5.4 to 13.5, p-values<0.05) (see Table 6).

Approximately three years after the intervention, all the children were assessed with a large battery of tests appropriate for their current age, which ranged from 4 to 7 years. At this long-term reassessment, we found significant differences in favor of children whose parents had originally received the PT intervention: their lexical and morpho-syntactic skills, as formally assessed through the PPVT, BNT, and TROG-2 tests, were significantly higher ($p<0.05$ by the Mann-Whitney U test; see Table 7). Differences in Peabody picture vocabulary and Boston Naming tests had a small effect size, with a probability of superiority of approximately 58%. The test for Reception of Grammar had a large effect size with a probability of 73.8% in favor of those children belonging to the PT group. No differences were recorded in listening skills (see Table 7).

Correlations between parents' and children's changes after the intervention

The CI-children's improvement in WC and SC, as measured through the MCDI, did not correlate with the mothers' and fathers' pre/post intervention changes. On the contrary, the improvement in WP was directly correlated with the mothers' GEN change (adjusted-rho=0.44, $p=0.02$). Correlations between the children's improvement in WP and pre/post intervention changes in mothers' OS and SENS score (adjusted-rho=0.35 and 0.34, respectively) were not significant ($p=0.08$ and 0.09, respectively). No correlation was found either between the CI-children's WP improvement and the fathers' changes on any of the tests, probably due to the small sample size (all p-values>0.1).

Lastly, a greater effect of the intervention on the CI-children's WP was observed when both parents took part in the PT. The mean pre/post-intervention change in children's WP score was 137±75 when both parents attended the PT (eight families) vs. 79±54 when only the mothers attended the PT (six families) ($p=0.02$).

Discussion

The role of the family is crucial to achieve optimal outcomes after cochlear implantation.; the degree of parental involvement is estimated to account for 35.2% of the variance in language (Moeller, 2000) and 20% of the variance in reading skills (Geers, 2003). During the last decade, parents' ability to establish responsive interactions in everyday life and to use adequate linguistic input has received increased attention because of the strong effect they have on language development in DHH children (DesJardin & Eisenberg, 2007).

In this context, the first aim of the present study was to assess the efficacy of a parent training program based on the ITTT model and adapted to the specific needs of DHH children including strategies by AVT, NAOA and Learn To Talk Around The Clock in terms of its ability to enhance parental responsive behaviors, improve linguistic development in children, and reduce levels of stress. The improvements in responsiveness observed in the parents after training were significantly greater than natural changes detected in the matched control group: both the mothers and fathers showed a greater sensitivity toward their children's communication; they tended to be more tuned in, had fewer difficulties in maintaining joint attention, and made more comments and expansions. Natural variations in the interaction style observed in the control group parents, although present, were less evident. These results confirmed the observations made by Glanemman et al. (2013) and provided new support to the idea that it is possible to induce significant changes in parent awareness and empowerment. Indeed, Glanemman et al. (2013) did not match their control groups with the study groups, thus introducing potential bias. The level of parental education, SES, and characteristics of the child, as well as the type of hearing device used, the hearing age, and the level of communication are all aspects that can influence the degree of responsiveness of both parents and children, and can cause variations in the ease with which they adapt to each other and attune their interactions (Geers et al., 2002; Glanemann et al., 2013). Because control group of the present study accounts for all of these factors, the present findings can be more strongly related to the effects of the parent training program. It is not possible to make a comparison with Harrigan and Nikolopoulus (2002) as they did not include a control group of untrained parents.

The second objective was to verify how the increased responsiveness of parents influenced the development of the listening and language skills of their children. Prior to parent training, there were no differences on the MCDI between the CI-children of the PT group and the control group. At the end of the group sessions, CI-children whose parents attended the PT showed a larger increase in word and sentence comprehension and word production, with significantly better performance than the children in the control group. Shortly after PT, a significant correlation was found between increasing in CI-children's word production and an improvement in the general aspects of the mothers' conversational behaviors, such as: their ability to use sentences of adequate length and complexity; to pause expectantly after speaking to encourage the child to respond; to speak to the child at an appropriate speed, intensity, and pitch and to use audition-maximizing techniques and appropriate gestures. This finding supported the hypothesis that attuned verbal inputs facilitate the early stages of word learning (Adamson, Bakeman, & Deckner, 2004; Baldwin, 1995; Tomasello & Farrar, 1986). Further, the word production score of children was higher when both parents, and not just the mothers, participated in the PT. Additionally, differences in language competence between the study and the control group were still present after three years: the CI-children of PT parents still demonstrated better lexical comprehension and production skills, and an even larger improvement in morpho-syntactic comprehension. Harrigan and Nikoulopolous (2002) observed a reemergence of nonresponsive behavior one year after the completion of PT. The consistency of the findings in the present study could be linked to the type of intervention, which includes a 6-month reinforcement and maintenance phase, during which parents could stabilize the new strategies they had learned.

Regarding listening skills, all CI-children showed excellent outcomes both in quiet and in noise, and their outcomes improved over time. The differences between PT and controls did not differ significantly. This last result reinforced the idea that listening skills alone, even when they are excellent, cannot guarantee children access to an adequate verbal language if this skill is not associated with a rich and tuned communicative interaction.

A further hypothesis was that the tests of language perception used in this as well as in previous studies were not sensitive enough to evaluate the correlation between language perception and language outcomes at a very young age (DesJardin, Ambrose, Martinez, & Eisenberg, 2009; Eisenberg, Shannon, Martinez, Wygonski, & Boothroyd, 2000; Newman & Chatterjee, 2013). The authors observed that speech recognition tests might not be nuanced enough to detect differences in more complex listening skills in children younger than 7-12 years. It will be necessary to develop new techniques and tools to better assess and track whether other emerging auditory abilities, such as auditory memory, have a greater impact on language development at such young ages.

The last area examined in this study was the indirect influence that a training program, that is based on communication empowerment, could exert on the perceived level of stress reported by parents of children with CI. No significant differences were found between the PT and control groups in PSI scores, either at the beginning and at the end of the habilitation process. Hintermair (2006) tested PSI in a large sample of hearing parents of DHH children, who showed how the degree of communicative competence influenced the parents' perception of stress. The author emphasized the importance of resource-oriented counselling and a support strategy in the early intervention because parental access to personal and social resources is associated with significantly less stress. The lack of statistical correlation found in the present study could be attributed to a number of reasons. The first one was the small study sample. Also, it must be considered that there are aspects of perceived stress which are influenced by contextual problems (financial status, marital relationships, parental personality, children's temperament, accessibility to support services, single parent, family size, etc.) and some that cannot be measured by the subscales of the PSI (personal sense of parenting, presence of depression, behavioral characteristics of children, etc.) (Cooper, McLanahan, Meadows, & Brooks-Gunn, 2008; Zaidman-Zait, 2008). Further, the PSI may not be the most sensitive instrument to detect significant changes regarding the impact of communication difficulties on perceived stress compared to specific measures for disability (Lederberg & Golbach, 2002; Quittner et al., 2010; Zaidman-Zait, 2008). Finally, one could argue that the pre- and post-treatment follow-up period was too short to observe significant differences in such profound aspects as the perceived stress level.

Limitations

Several limitations should be considered in relation to current results. First, there were a small number of children and families involved in the study and we hope to increase the study group to confirm the findings. Secondly, it is possible that, despite our attempts to minimize limitations through matching demographic, audiological, linguistic, and parent's responsiveness characteristics before training began, we introduced unintentional biases when we planned the study and chose not to randomly assign parents and children to the PT or to the control group. Third, families that agreed to participate in PT may have been more motivated and compliant than others used as a control group, as one might infer from the lack of dropouts during the program. Finally, individual sessions at home, although desirable as they allow clinicians to help families in their real-life context, were not always feasible due to logistical problems (family distance from the CI center), economic costs, and time spent.

Clinical Implications and Future Directions

These results have several implications in planning habilitation intervention strategies for CI-children and their families.

Results suggest that experts can help parents to learn strategies that are useful to enhance the linguistic development of their CI-children. It is possible to modify parents' communicative behaviors and a PT program which combines group and individual sessions could reinforce these new techniques and behaviors. During the group sessions, specialists present strategies and the parents and specialists both engage in discussions. In the individual sessions, the specialists were able to indirectly guide and assist parents towards awareness of their communication style and the effects it exerts on the development of children's language. A future study that utilizes a randomized clinical trial could provide more validity to these results. It would also be helpful to increase the sample size in order to verify the effectiveness of the PT and the compliance of families who come from multicultural environments or with more disadvantaged situations and low SES.

A further direction of the study should be to include other caregivers, such us teachers, in order to improve communication in all contexts in which children can benefit from interactions with adults.

Finally, specialists could implement the use of tele-practice (McCarthy, Muñoz, & White, 2010) to overcome the challenge of organizing individual sessions at the families' homes and to include families who live far away from habilitation services and would not otherwise be able to benefit from the program due to logistical problems.

Conclusion

Parents' responsiveness and sensitivity played a fundamental role in the promotion of linguistic, socio-emotional, and cognitive development in children with DHH. The differences in language skills observed between the PT group children and the control group showed a significant correlation with the shared language and the quality of the interaction with their parents. The experience gained by parents during the training seems to represent a solid basis for their children's language learning. The use of ITTT principles, reinforced by information and strategies specific for DHH children, seems to be effective in achieving this goal. Further studies are needed to expand our knowledge in this field and to produce more generalizable results.

Chapter 3. Cochlear implantation in infancy: the importance of sound in boosting human brain architecture

3.1 Emotional valence of an early auditory scaffolding

Emotional competence can be defined as the capacity to reach one's goals after an emotion-eliciting encounter. In this sense, it represents a crucial element for self-efficacy (Saarni, 1999). Emotional competence is based on awareness of individual emotions and their influence on others: it encompasses the ability to understand his own personal emotions which precedes the ability to evaluate and understand others' emotions. It is the core for empathy: recognizing one's own emotions opens to the possibility of responding properly to the emotions that other people experience. Without knowing one's own emotions, it is difficult to feel empathy for another.

Saarni (1999) highlighted eight skills as the components of emotional competence: (1) awareness of one's own emotions, (2) comprehension of others' emotions, (3) use of the vocabulary of emotion and expressions, (4) empathic involvement, (5) ability to differentiate subjective emotional experience from external emotion expression, (6) coping adaptively with aversive emotions and distressing circumstances, (7) awareness of emotional communication within relationships, and (8) emotional self-efficacy.

Lau (2006) identified three building-block for emotional competence: the ability to identify personal and others' emotions; the ability to communicate emotions to others and the ability to cope with negative emotions.

According to Susanne Denham (Denham, 1998), the construct of emotional competence embraces three broad emotional abilities:

1. Expression
2. Comprehension
3. Regulation

Emotional expression means using gestures to express non-verbal emotional messages, demonstrate empathic involvement, manifest social emotions, be aware that it is possible to control the manifest expression of socially disapproved emotions.

Emotional comprehension, on the other hand, allows us to discern our own and others' emotional states and to use an adequate emotional vocabulary.

Finally, emotional regulation deals with facing and managing both negative and positive emotions or the situations that arouse them.

Emotional comprehension and emotional competence are therefore not strictly synonymous; rather, emotional comprehension constitutes an aspect of the more general development of emotional competence and it is limited to the ability to identify, predict and explain one's own and others' emotions (Harris, 1989).

According to Harris (1999) early caregiver sensitivity to emotional states is responsible for individual differences in children's understanding of emotion. Moreover, the parental ways and in which feelings are put into words may lead to variation in children understanding of emotions. A key element for emotional development is parental responsiveness, the ability of caregiver to “tune in” to their children, to recognize their child’s communicative attempts, their emotional states and provide contingent responses (Suskind, Suskind, & Lewinter-Suskind, 2015). So, if children’s emotions are not attended with support and care, dysfunctional coping strategies can emerge, leading to ineffective emotional competence and regulation, or in other cases, to avoidance of recognition and expression of emotions (Colle & Del Giudice, 2011).

As a matter of fact, since her first weeks of life, the child is an "active listener", in particular towards human sounds (Moore & Lintichum, 2007). This early innate ability to perceive the environmental sounds is basically essential for development of language: the acquisition of spoken language requires auditory input and interaction with the environment. At birth, human cochlea is actually mature, but it begins to function from the 25th week after conception; the development of the afferent auditory path begins before cochlear functioning is fully stabilized and maturation of nervous system goes on well after birth. Starting from the 1980s, numerous psychological researches have shown that newborn babies show a preference for the mother's voice at birth. The historical research of DeCasper and Fifer (1980) showed that the infant after a few hours of birth shows preference for mother’s voice over that of other women. In another experiment, De Casper & Spence (1986) observed the ability of newborns to discriminate between two different fairy tales, showing a preference for the story told them every day for ten minutes in the last trimester of pregnancy. The prenatal sound exposure therefore already seems to establish the basic elements for language learning: for example, it has been observed that the sound spectrogram of the cry of premature babies already contains the specific vocal characteristics of the mother's voice.

Between six and nine months, listening skills become more and more mature and specialized: child begins to distinguish the sounds of language from different speakers (Kuhn,

1979) with an increasing preference for the sounds of the native language and a substantial decreased ability to distinguish and encode phonemes not belonging to native linguistic environment (Jusczyk et al., 1993).

In a key study Saffran (1996) found that six-month-old babies were able to pay attention to phonological and auditory stimuli according to statistics relating to environmental linguistic phonemes. This early preference for his own mother tongue phonemes is based on reference to "statistical properties" of the sound environment via a so called "probabilistic learning" (Marcelli & Cohen, 2009). Furthermore, the growing brain perceptual system preference for his own mother tongue is related to an "affective" learning, also called "attraction effect". In this sense, an important role is played by the affective involvement of the caregivers, both for the acoustic and emotional characteristics of maternal baby talking, both for the experience of a steady and predictable emotional scaffolding.

Even before birth, presence of cochlear dysfunction entails detrimental effects on the functional integrity of the auditory pathways, with possible consequences also on higher cognitive functions and systems that rely on those sensory inputs, including emotional comprehension skills.

As described in next paragraph, a severe-profound deafness might significantly alter the complex process that brings children to develop a mature emotional comprehension.

Cochlear implant allows deaf children to develop better auditory and linguistic skills: in next paragraph the possible impact of an early cochlear implantation in emotional development of congenital deaf children and a number of audiological, linguistic and psychological factors that may affect emotional skills are discussed.

3.1.1 Level of emotion comprehension in children with mid to long term cochlear implant use: How basic and more complex emotion recognition relates to language and age at implantation

Abstract

Objectives: The current study was designed with three main aims: To document the level of emotional comprehension skills, from basic to more complex ones, reached by a wide sample of cochlear implant (CI) deaf children with at least 36 months of device use; To investigate subjective and audiological factors that can affect their emotional development; To identify, if present, a “critical age”, in which early intervention might positively affect adequate emotional competence development.

Design: This is an observational cohort study. Children with congenital severe/profound deafness were selected based on: aged by 4-11 years, minimum of 36 months of CI use, Italian as the primary language in the family; normal cognitive level and absence of associated disorders or socio-economic difficulties. Audiological characteristics and language development were assessed throughout standardized tests, to measure speech perception in quiet, lexical comprehension and production. The development of emotions' understanding was assessed using the Test of Emotion Comprehension (TEC) of Pons and Harris, a hierarchical developmental model, where emotion comprehension is organized in three Stages (external, mental and reflective). Statistical analysis was accomplished via the Spearman Rank Correlation Coefficient, to study the relationship between the personal and audiological characteristics; a multivariate linear regression analysis was carried out to find which variables were better associated with the standardized TEC values; a chi-squared test with Yate's continuity correction and Mann-Whitney *U* test were used to account for differences between continuous variables and proportions.

Results: 72 children (40 females, 32 males) with a mean age of 8.1 years were included. At TEC score, 57 children showed normal range performances (79.17% of recipients) and 15 fell below average (20.83% of recipients). The 16.63% of older subjects (range of age 8-12 years) didn't master the Stage 3 (reflective), which is normally acquired by 8 years of age and failed 2 or all the 3 items of this component. Subjects implanted within 18 months of age had better emotion comprehension skills. TEC results were also positively correlated with an early diagnosis, a longer implant use, better auditory skills and higher scores on lexical and morphosyntactic tests. On the contrary, it was negatively correlated with the presence of siblings and the order of birth. The gender, the side and the severity of deafness, type of implant and strategy were not correlated.

Conclusions: Early implanted children have more chance to develop adequate emotion comprehension, especially when the complex aspects are included, due to the very strong link between listening and language skills and emotional development. Furthermore, longer CI auditory experience along with early intervention allows an adequate communication development which positively influences the acquisition of such competencies.

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Introduction

Emotion Comprehension refers to the ability to identify, predict and explain one's own and others emotional experience (Denham et al., 1990; Pons, Harris, & de Rosnay, 2004; Southam-Gerow & Kendal, 2002). A mature understanding of emotion is critical to children's social development as it affects specific characteristics of the individual's identity, such as self-esteem, self-concept, and self-identity (Gimenez-Dasì, 2013), and influences the ability to show empathy and establish significant relationships with others ((Denham et al., 1990). The strong link existing between emotion comprehension and social competence from a very young age through adolescence is well documented by studies on normally hearing children (NH) (Custrini & Feldman, 1989; Denham, 1998; Denham et al., 2003; Mostow et al., 2002).

The level of emotion comprehension in preschool and primary-school children has been shown to deeply affect the quality of prosocial behavior with classmates and teachers and the quality of shared play, showing a positive correlation with reductions in behavioural problems (Pons, Harris, & de Rosnay, 2004; Cassidy et al., 1992; Dunn & Cutting, 1999). Children who comprehend and appropriately manage emotions are also more socially skilled, achieving greater integration and social acceptance (Denham et al.,1990; Edwards, Manstead, & Mac Donald, 1984) and are more competent in resolving interpersonal conflicts (Herrera & Dunn, 1997; McDowell, O'Neil, & Parke, 2000).

Emotions occur in the context of ongoing social interactions and relationships, and interact with the internal cognitive, motivational, and physiological components that characterize each individual (Gimenez-Dasì, 2013), so their maturation is a complex and gradual process that starts in early years and continues until adulthood. Children are able to discriminate facial expressions delivering happiness and surprise by the age of three months (Young-Browne, Rosenfeld & Horowitz,1997), anger by four months (Barrera & Maurer, 1981), and fear by seven months (Kotsoni, de Haan, Johnson, 2001). By seven months they are also able to associate the tone of voice to the corresponding emotional facial expression (Soken & Pick, 1991. Infants also use other people's emotion to gather information about objects or events (Walden & Ogan, 1988): by the age of 12 months they use this mechanism to determine whether to approach or avoid a novel object (Klinnert et al., 1986) and then by the age of 18 months to infer other people's desires even when in conflict with their own (Repacholi & Gopnik,1997). By the age of two years, children progress in their comprehension of nature, causes, effects, and regulation of their own and others' emotions, and achieve a series of skills that develop throughout early childhood until twelve years of age. These skills have been described by Pons and co-authors (Pons et al., 2004; Harris, 1989; Pons & Harris,2005) in a developmental model, where they are organized in nine hierarchical components:

Component I: Recognition. Children aged approximately three e four years are able to recognize and name basic emotions (happiness, sadness, anger and fear) on the basis of facial expressions;

Component II: External Causes. Children understand that external causes can influence their interior emotional status; for example, they know that the loss of a toy usually makes a child sad and they can anticipate the happiness another feels when he re-ceives a gift;

Component III: Desires. At around 4-5 years, children manifest understanding that different desires can cause different emotions; as a consequence, at this age a child can comprehend that two people are able to experience different emotions in the same situation due to different inclinations;

Component IV: Belief. In the same period, children show an emerging comprehension that their own beliefs, whether true or contrary to reality (false-beliefs), can influence emotional reactions;

Component V: Memory. Children aged 3-6 years begin to show evidence that they understand the influence of reminders on intensity of emotions: in this sense, children start realizing that time reduces the intensity of emotion and that some past emotions can be reactivated by some aspects of a present situation;

Component VI: Regulation. A more advanced comprehension of emotions emerges approximately at 8 years, when children begin to use psychological strategies (such as, negation, distraction) to regulate and face negative emotions, whereas at earlier ages children refer mostly to material and behavioural strategies;

Component VII: Hiding. By six years, children typically demonstrate the ability to differentiate between apparent and real emotions. In particular, they show both the ability to hide their own emotions and to identify dissimulation in others. In this sense, they understand that people may internally experience emotions that differ from those they intentionally display;

Component VIII: Mixed Emotions. Moreover, by 8-9 years, children show the ability to comprehend that a situation can elicit mixed and contradictory emotions, for example experiencing happiness and fear simultaneously;

Component IX: Moral Emotions. Furthermore, at the same age, many children manifest appreciation of morally based emotions and ethical dimensions of feelings. In fact, even if children can already express guilt at the preschool age, it is only in middle childhood that they systematically recognize when a person might be inclined to feel guilty, showing a lag between the expression and the attribution of moral emotions.

Components VI, VIII, and IX are considered to be the most advanced skills in emotion comprehension. Affective interchanges and communicative sharing, in the particular cultural settings where they occur, such as conversations with parents at home or with peers during free play (Hepach, G. Westermann, 2013), have been seen as the two most important factors in determining correct emotional development (Montague & Walker-Andrew, 2002; Bosacki &Moore, 2004). The family context is where initial exposure to other people's expressions occurs and where infants can participate in affective interchanges and observe caregiver's responses to themselves and/or to others (Montague & Walker-Andrew, 2002). Parents begin soon after birth to interact with their newborn child, giving him or her care and love, interpreting each infant state (physiologic and affective needs, wishes, facial expressions), and labelling them using a mixed code that is made of touch, gazes, facial expressions, and language sharing (Landry, Smith, & Swank, 2007). Sensitive and responsive parenting facilitates the organization of infants' physiological systems to achieve the internal regulation necessary to support more complex social, emotional, and cognitive development (Landry, Smith, & Swank. 2007; Panksepp, 2001). In this process, language is the connecting link which represents and organizes emotional meanings.

Language supports emotional exchanges and helps children to link a particular expression, action, or event to its external referent as well as labeling the resulting elicited emotion. The temporal synchrony between voices (vocal intonation), facial expressions, language, and emotional elicitors (object, event) have been shown to be necessary for younger infants to learn and discriminate emotions (Flom & Bahrick, 2007). NH children can directly listen to adult's interpretations relating to a specific emotion and at the same time observe the social context in which everyday interactions and conversations occur.

Words or labels play a large role in the development of the child's conceptualization of emotions (Saarni, 1999). Parent's use of a richer emotional language seems a significant predictor of future emotional skills in children (Dunn, Brown, & Beardsall , 1991; Denham & Auerbach, 1995). Both semantic-lexical abilities and syntactic competence have been found to be strongly and positively correlated with the ability to comprehend emotions (Bosacki & Moore, 2004). Contingent responding, that is, warm acceptance of infant's needs and interests, is thought to support the infant's self-regulatory skills by favoring the development of mechanisms for coping with stress and novelty, along with trust and bonds with the caregiver through the process of internalization (Landry, Smith, & Swank. 2007) .

Severe-Profound deafness can alter the complex process that brings children to develop a mature emotional competence for two main reasons: the lack of a full sharing of verbal communication (Ludlow et al., 2010) and the qualitative-quantitative alteration in parent-child interactions (Cole & Flexer, 2007). Studies on deaf populations with traditional hearing aids (HA) have shown delayed development (Ludlow et al., 2010; Most & Michaelis, 2012; Rieffe, 2012), “with a narrower and less flexible perception of emotional situations due to restricted opportunities to learn from their own and others' experiences via the auditory channel” (Ziv, Most & Cohen, 2013).

With the advent of cochlear implantation, which allows children who are severely or profoundly deaf to develop better auditory skills and offers wider opportunities to gain adequate language competence (van Wieringen & Wouters, 2015), a new interest in the assessment of emotion comprehension in deaf population has encouraged studies on the impact of CI on child development.

Most of these studies were designed to investigate the development of basic competencies (Most & Michaelis, 2012; Ziv, Most & Cohen, 2013; Wang et al., 2011; Wiefferink et al., 2015) and only two of them focusing also on more complex skills (Wiefferink et al., 2015; Ketelaar et al., 2015), with apparently contrasting results. Most and Michaelis (2012) studied both visual and auditory perceptions of emotion in CI and NH preschoolers. Wang et al.'s study (2011) considered visual perception of expressions of happiness, sadness, anger, and fear, comparing CI and HA children with age matched NH children. Both studies found significant differences in outcomes between groups, with worst performances observed in HA and implanted users.

On the contrary, Hopyan-Mysakyan et al. (2009), while finding significant differences between CI and NH children in perceiving the affective speech prosody task, reported no overall significant group differences concerning the visual assessment of the facial affect tasks. Most and Aviner (2009), when assessing emotion comprehension in auditory-visual mode, found significant differences comparing CI with NH children for identification of surprise, but not for the identification of happiness, anger, sadness, fear, and disgust. Wiefferink et al. (2015) and Ziv et al. (2013) extended the analysis including emotion attribution in situational contexts and the ability to infer affective perspective from stories. Their outcomes were also contrasting since Ziv et al. (2013) found that CI children exhibited comparable abilities with NH peers in understanding emotions of others from stories, even when they differed from their own, whilst Wiefferink et al. (2015) reported that CI children performed worse than NH controls. Finally, Wiefferink et al. (2015) and Ketelaar et al. (2015) analyzed more complex components of emotion comprehension, assessing the ability to understand a situation which can elicit mixed and contradictory emotions, and emotions based on shared moral rules, but included in their samples preschool CI children only. Their performance was significantly lower compared to NH peers, but the authors did not investigate the skills of school age CI children and it is not possible to know if with increasing age and auditory experience the gap will be filled.

None of these studies investigated whole emotional skills in the same sample, offering only partial insights on individual components. Furthermore, all of them showed substantial differences in study group composition (length of auditory experience with CI, age at test, and mode of communication) and are lacking in long-term observation of the effects of late progresses in language competencies on the development of emotional comprehension. These differences might have also contributed to apparently contrasting study outcomes.

The current observational cohort study was designed with three main aims:

- to document for the first time the level of emotional comprehension skills, from basic to more complex competencies, reached by a wide sample of cochlear implanted deaf children with at least 36 months of device use, through a standardized test;

- to investigate subjective and audiological factors that can affect their emotional development;
- to identify if there exists a “critical age” in which early intervention might positively affect adequate development of emotional competence.

It might be expected that, due to the new listening possibilities offered by cochlear implants and a longer auditory experience, most deaf children could gain adequate emotion comprehension skills. In this respect, early implantation by assuring early access to language could be one of the main predictors for higher performance in congenitally deaf children, providing early maturation of basic competencies and offering opportunities to develop more complex skills, such as understanding of moral rules. Finally, although an increment in chronological age and increased cochlear implant use could positively affect basic competencies such as emotion identification skills, it might be expected that age at implant would have an effect on outcomes. Similarly, as for language skills (Fulcher, et al., 2012), children implanted after 18 months of age could have less chances of reaching full maturation of more complex emotional competencies.

Materials and methods

Study design

The present research is based on an observational cohort study according to the rules of the STrengthening the Reporting of OBservational studies in Epidemiology (STROBE) statement (2015). The protocol was approved by the local ethics committees of the two Italian Cochlear Implant Centers that cooperated for the study's implementation and realization. The recruited families gave written informed consent for their own child's assessment before commencing any study-related procedure.

Participants

Children with congenital severe/profound deafness (Pure Tone Average in the better ear 70 dB HL for 500e4000 Hz), aged 4-12 years at the time of enrollment, were included. The choice of age was related to the reference age included and standardized in the emotion comprehension test. Additional inclusion criteria were a minimum of 36 months of CI use which represents a period in which early intervention allows an adequate communication development (Fulcher, et al., 2012; Nicholas & Geers, 2012); Italian as the primary language used in the family; normal cognitive level; and absence of associated disorders or socio-economic difficulties.

Normal cognitive level was established as being > 25 percentile at Raven's Coloured Progressive Matrices-CPM (Raven, 1965).

The absence of associated disorders was verified by clinical history. Normal socio-economic status was defined as 13 years of mother/father schooling (high school level) and annual family economic income 29.956 euros (Italian National Institute of Statistics, ISTAT).

Information about children (gender and birth order), audiological aspects (deafness severity, age at diagnosis, age at implantation, type of used cochlear implant, speech processing strategy, monaural/binaural condition), and type of rehabilitation (traditional aural/oral therapy, auditory verbal therapy or oral sign language therapy) were collected for each recipient and were used for the analysis of factors influencing emotion comprehension skills.

Auditory skills assessment

Speech recognition in quiet conditions was assessed using standard Italian phonetically balanced bisyllabic words for paediatric populations (Cutugno, Prosser &Turrini, 2000). A 10 items list was preceded by a practice list. Items were administered in a soundproof room, via a loudspeaker placed at 1 m distance from a table where the child was sitting next to a speech therapist. Stimuli were presented at 65 dB SPL 0 azimuth and score was calculated as % of words correctly repeated.

Cognitive skills assessment

Subjects with cognitive delay were excluded from the study because individual differences in emotion comprehension skills may also depend on different levels of processing skills (Albanese et al., 2010). Non-verbal cognitive abilities were measured via Raven's Coloured Progressive Matrices-CMP (Raven, 1965). CPM is a culture free test, widely used to measure non-verbal cognitive abilities in children aged 4-11 years. In particular, it is composed of 36 items that are arranged to assess cognitive development and mental ability up to the stage where a child is sufficiently able to reason by analogy as the prevalent method of inference.

Language skills assessment

Language abilities were assessed using three Italian Standardized Language tests. Lexical comprehension was assessed using the Italian version of the Peabody Picture Vocabulary Test-PPVT, on which normal standardized scores range between 85 and 115 (Dunn & Dunn, 1981). Lexical production was measured using the Italian version of the Boston Naming Test-BNT (Riva, Nichelli & Devoti, 2000), adapted for school children and normal adults. The authors provided mean scores and standard deviations for a sample of 160 school children, so z-scores were calculated, and $z > 1$ was considered normal. Morphosyntactic comprehension assessment was undertaken using the Italian version of the Test for Reception of Grammar 2 (TROG-2) and referring to its standard normalized scores: these range from 85, that is the standard score corresponding to the 16° c, given by the TROG-2 manual as -1 sd below the normative sample average, and 115 that corresponds to 84° c, given by the TROG-2 manual as 1 sd above the average; performance below 85 are considered pathological; performances equal or above 115 are considered higher than the average (Bishop, 2003).

Emotion comprehension assessment

The development of emotional understanding was assessed using the Test of Emotion Comprehension (TEC) of Pons and Harris (2000). The TEC consists of a picture book showing a sequence of cartoons presented in a fixed order of increasing difficulty, and has two versions, one for males and one for females. The TEC includes 23 tables, divided into nine blocks assessing the nine components of emotion comprehension, as decrypted in the Pons and Harris model to explain emotion comprehension development in children by 3 years to 11 years of age.

The Italian version of the original British test (Albanese & Molina, 2008) was standardized on a sample of 967 children from different parts of the country (Northern, Center, and Southern parts) so it is considered highly representative of the whole population. The TEC shows a good reliability coefficient (Rho = 0.79), with a test-retest reliability equal to r ¼ 0.78. Furthermore, the test shows correlations with language ability (r =0.59) and cognitive development tasks (r = 0.60). The different components of the TEC are scalable (Index of consistency I = 0.676) and the scale is valid (Coefficient of reproducibility R ¼ 0.904) (Albanese &Molina, 2008).

The administration procedure is divided into two parts.

The first part (Tables 1-5), that assesses recognition of emotion on the basis of facial expressions (Component I) is a traditional task of identification of basic emotional facial expressions (happiness, sadness, fear, and anger): for each table, the child is presented 4 images representing them and is requested to make a direct association between the emotion named by the examiner and the corresponding image.

The second part (Tables 6-23), that assesses all the other components of the Pons and Harris model (2000), is presented as an understanding emotion task using situational contexts to elicit basic emotions: the examiner reads short stories to the child while looking at a cartoon drawing, and then asks him to indicate the correct emotional expression which is expected to be elicited by the plot. These drawings present an unvarying structure characterized by a simple cartoon strip on the top of each page, and four faces expressing different emotional cues in the bottom part. In each table the target expression is presented together with the neutral expression and all the other emotions in a random order.

In detail, Tables 6 to 10 assess children's comprehension of external causes of emotion (Component II): e.g., understanding that a child will be happy if given a gift.

Tables 11-12 assess understanding of desire-based emotions (Component III): i.e., attribution of different emotions to two characters with opposite desires about the same situation.

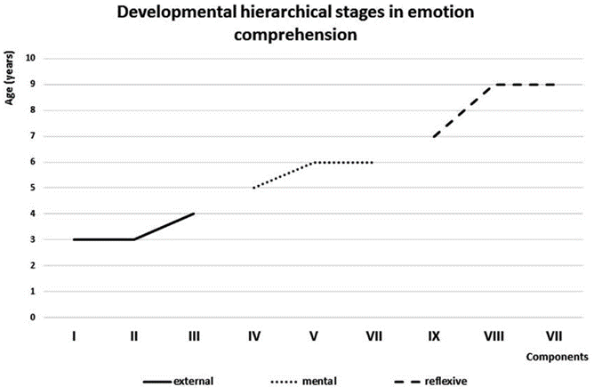
Tables 13 to 18 evaluate in a linked narrative format Components IV, V, and VI. In particular, the comprehension of belief-based emotion (Component IV) is assessed by item 13, in which children are asked to attribute an emotion to a character holding a false belief. Then, the narrative goes on through Tables 14-17, assessing understanding of the influence of a reminder on pre-sent emotional state (Component V) and finishes with table 18 that evaluates the regulation and control of emotions (Component VI): the child is asked to choose the best way between four possible regulation strategies: a deceptive strategy, a behavioral strategy, a psychological strategy, or no strategy. Table 19 assesses if there is comprehension that one can hide or dissimulate an underlying emotion (Component VII): e.g., understanding that a person can smile in order to hide her distress from another.

Table 20 addresses children's understanding of mixed emotion (Component VIII) through a narrative about ambivalent feelings in the protagonist (e.g., sadness and fear).

Finally, Tables 21-23 assess children's understanding of moral emotions (Component IX), meaning the ability to comprehend that guilty or negative feelings can be elicited by misbehaviours or immoral actions, whereas praiseworthy actions induce positive emotions.

Answers are non-verbal, and children are requested to point to the correct picture right after the item is presented. All tasks were presented to each child; 1 point was assigned for each component correctly answered (for a total maximum score of 9 points). The total maximum raw score by each child was converted into z-scores according to the normative data given by the Italian version of TEC handbook (Albanese &Molina, 2008).

Although there is substantial individual variability in children's responses at any age, Pons et al. (2004) identified a hierarchical developmental pattern, with three progressive stages. Stage 1 (External) is when, as in early childhood, children focus on visible and public aspects of emotions: it is defined by correct answers to Components I (identification), II (external causes), and V (memory). Stage 2 (Mental) is when children show an increasing knowledge of mental aspects, such as desires and beliefs: it is defined by correct answers to components III (desires), IV (belief), and VII (hiding), and is usually reached by normally developing children between 4 and 7 years of age. Stage 3 (Reflective) is when children show an increasing awareness of psychological and reflective aspects of emotion: it is defined by correct answers to components VI (regulation), VIII (mixed emotions), and IX (moral emotions), and is usually reached in normally developing children by 8 years of age (Fig. 3). In each stage a child can obtain a raw score of 3, if he/she answers correctly to all the three components included into the stage. As no normative data are given for each Stage (External, Mental and Reflexive), in the statistical evaluation the raw scores were used for the analysis.



This graph is a scheme of the hierarchical developmental pattern of emotion comprehension, organized in three progressive stages, related to the age where normal hearing children usually develop each competence (Pons et al., 2004).

Assessment procedure

Assessment was carried out in two different sessions: one for audiological and linguistic assessment and the other for cognitive and emotion tasks administration. Each session lasted approximately 1.5 h. The sessions were a few days apart from each other.

Children were tested individually in a quiet room, by two female speech therapists, for the language assessment, and by a female psychologist, for cognitive and emotion comprehension evaluation. All children communicated orally, so all tests were conducted using spoken language, using live voice at a level of conversational speech. Items of linguistic, cognitive and emotion comprehension tasks were presented in a visual-auditory modality. Each child was seated at a table, that was approximatively 50 cm large, and the examiner was seated in front of him/her.

Statistical analysis

Data are presented as means (standard deviation, sd) or median [range] for continuous variables and as proportions for categorical variables, as appropriate.

The chi-squared test with Yate's continuity correction and Mann-Whitney *U* test were used to account for differences between continuous variables and proportions, respectively. The relationships between the personal and audiological characteristics of study sample, language skills, and the outcome measures were investigated by the Spearman Rank Correlation Coefficient. Standardized z-score values according to normative Italian populations were used for TEC total score (Albanese &Molina, 2008).

Since standardized values were not available for the partial scores for the three developmental Stages (external, mental and reflective), Spearman Rank Correlation Coefficients were carried out by using raw subtest scores adjusting them for age at assessment.

The normal distribution of total TEC z-score was verified by means of the Shapiro-Wilk test, resulting in a normal distribution (p = 0.19), therefore a multivariate linear regression analysis was carried out to find which personal, audiological, and linguistic variables (independent variables) were better associated with the standardized TEC value; gender and age at study evaluation were included in each model as covariates.

For the multivariate analysis, all variables of interest were added in models in a stepwise fashion, and interactions terms were tested where appropriate. In each subsequent step, the regression equations comprised those predictors reaching specific thresholds of F- and p-values (for predictor inclusion: $F \geq 1$ and $p \leq 0.05$; for exclusion: $F<1$ and $p > 0.05$).

To identify if there was a “critical age” in which cochlear implantation allowed the development of adequate emotional competencies, children were divided into five groups, according to the age at implantation, with a narrower window in the first two years, where most of the attention is placed for implications for language acquisition (Houston et al., 2012), and wider windows for the following ages at implantation: ≤12 months group, 13-18 months group, 19-24 months group, 25-36 months group, ≥37 months group. Multi-group comparisons were performed by means of one-way analysis of variance (ANOVA) with the LSD procedure as a post-hoc test.

All p-values less than 0.05 in either direction were considered as significant. Analyses were carried out using a PC version of Statistical Package for Social Sciences 16.0 (SPSS, Chicago, IL, USA).

Results

Subjects

72 children (40 females, 32 males) with a mean age of 8.1 (2.3) years (interval from 4 to 12 years) were included. They came from different Regions of Italy (North, Centre, and South) and were enrolled in two Cochlear Implant Center.

All children had congenital sensorineural hearing loss, which was profound in 59 and severe in 13 of them. Aetiology was: unknown (34, Connexin 26 mutation (36), ototoxicity (1) cytomegalovirus infection (1). Mean chronological age at diagnosis was 15.2 months (range 1e38) while mean age at implantation was 22.1 months (range 7-48). The mean time of device use at assessment was 6.1 years (range 3.7-10.6 years).

Forty-nine recipients were implanted with Cochlear devices and used Freedom or CP810 speech processors, fitted with ACE strategy; 23 received Advanced Bionics devices and used Auria, Harmony or Neptune speech processors, with Hi-Res 120 strategy. 16 children received simultaneous bilateral cochlear implants, 18 adopted bimodal hearing (CI and controlateral HA) while 38 where unilateral CI users.

Concerning rehabilitation mode, 50 children participated in a Auditory-Verbal Therapy (AVT), 17 in an Auditory-Oral program (AOP) and five in oral rehabilitation programs with sign language used as support for communication (ISL support). Nevertheless, at assessment, none of these children used sign language at school, neither at home, and all used oral verbal communication only. All the children were included in normal mainstreamed schools, with a support teacher, according to the normal legislative procedure of the ministry of education.

The sample average normalized score at CPM - measuring non-verbal intelligent skills was 83.2 (range 29-100).

Average disyllabic words recognition percentage was 89.03% (range 60-100). Standard PPVT average score was 82.7 (range 54-111), with 63.89% of children fell within the normal range for lexical comprehension. The average z-score at BNT was -0.34 (range -5-4), and 68% of children fell within the normal range for lexical production.

The average standard score at TROG-2 was 96.9 (range 55-127), and 65.3% of children fell within the normal range for morpho-syntactic comprehension.

Emotion comprehension skills

The mean standardized TEC value (z-scores) of the study sample was 0.189 (interval from 3.310 to 2.200), with 57 children (79.17% of recipients) showing normal range performances, with z-scores > -1 and 15 (20.83% of recipients) falling below average (z-scores ranging from -1.000 to -3.310). Table x shows the mean general level of emotion understanding by age in the study CI group and Italian Normative Sample (Albanese &Molina, 2008) and shows a fairly linear improvement in general level of emotion understanding as a function of age, in CI children as in NH children. Younger CI children, from 4 to 7 years of age, have higher mean total raw score than the corresponding NH peers of the Italian Normative Sample. Mean total raw scores became similar between older CI children and Italian NH children, from 8 to 11 years. The two 12 years old CI children, both with an age at implantation >24 months, had instead a lower raw score.

	Study sample			Italian normative values		
Age at evaluation	n	Mean (SD)	Range	N	Mean (SD)	Range
3 years	0	-	-	110	2.1 (1.3)	0-7
4 years	4	6.3 (1.5)	4-7	113	3.2 (1.7)	0-7
5 years	10	5.7 (1.9)	2-8	109	4.6 (1.8)	0-8
6 years	14	6.0 (1.0)	4-7	107	5.6 (1.4)	2-8
7 years	9	7.2 (1.5)	5-9	107	6.3 (1.4)	2-9
8 years	6	7.2 (1.8)	5-9	106	7.1 (1.3)	4-9
9 years	10	7.4 (1.6)	4-9	104	7.7 (1.2)	4-9
10 years	8	7.6 (1.3)	6-9	104	7.9 (1.2)	2-9
11 years	9	7.9 (0.8)	7-9	107	8.0 (1.0)	4-9
12 years	2	6.5 (0.7)	6-7	-	-	-
Total	72	6.8 (1.6)	2-9	967	5.8 (2.5)	0-9

Mean raw scores (and standard deviation) by age of the general level of Emotion Comprehension (TEC), measure in CI subjects and in the Italian NH normative samples

In CI subjects, as in NH children, the percentage of correct answers increases with age, although the improvement was much slower for some components than for others (see supplementary table I: supplementary data related to this article can be found at http://dx.doi.org/10.1016/j.ijporl.2016.06.033)

The raw scores of each of the TEC Stages were as follows: Stage 1 (External), median = 3 (range 1-3); Stage 2 (Mental), median = 3 (range 0-3); Stage 3 (Reflective), median =2 (range 0-3) (Table x). Regarding the 15 CI children falling out of the normal range, 3 (4.2%) were younger (age 4-6 years) and failed reaching the Stage 2 (mental), and 12 (16.63%) were older CI children (8-12 years) and didn't mastered the Stage 3 (reflective) which is normally acquired by 8 years of age. In details, all of them failed the item measuring the capacity to understand morality (Component IX); 10 failed in passing from more behavioral strategies for emotional control to psychological strategies (Component VI); 7 showed difficulty in understanding that a person may have multiple or even contradictory (ambivalent) emotional responses to a given situation (Component VIII).

Factors that affect emotional comprehension skills

Of all subjective and audiological variables that may affect emotional comprehension performance, the gender, the severity of deafness, type, side of implant and coding strategy did not affect the total TEC scores (all p values > 0.1). By contrast, the presence of siblings and the order of birth seemed to affect performance: the second and third-born CI children scored worse than first-borns and only children, and this was true even after adjusting analysis for age at study entry (all p-values<0.01). A statistical trend for better normalized TEC total scores and for partial raw scores in items belonging to the external stage component were found in patients with binaural listening condition ($p = 0.062$ and $p = 0.053$, respectively). However, these figures did not survive after adjusting analysis for age at study entry.

A younger age at implant ($r = 0.58$, $p < 0.001$), a younger age at diagnosis ($r =0.57$, $p < 0.001$), a longer time of implant use ($r = 0.26$, $p = 0.03$) and Bysillabic Word Recognition ($r =0.55$, $p < 0.001$) were related to better TEC scores, especially in terms of achievement of the Stage 3 (Reflective) (ranging from 0.53 to 0.43; all p values < 0.02) (see Table 3: it can be found at http://dx.doi.org/10.1016/j.ijporl.2016.06.033). We also found significant correlations between linguistic skills and TEC score, being Stage 2 (Mental) and Stage 3 (Reflective) closely related to higher scores on lexical (PPVT) and morphosyntactic (TROG-2) tests (see also Table 3).

The multivariate linear regression analysis showed that the normalized TEC score was associated with lexical production, assessed through BNT ($p < 0.001$), Bysillabic Words Recognition ($p =0.001$) and age at implant ($p =0.033$) (see Table 4 at http://dx.doi.org/10.1016/j.ijporl.2016.06.033). These three variables accounted for about 50% of the explained variability in this measure (adjusted R-squared 0.495). The regression equation to estimate the standardized TEC z-score was: -10.485 (constant) + 0.270 multiplied by normalized score at BNT + 0.025 multiplied by percentage of correct items at bysillabic words' recognition test -0.022 multiplied by age at implant (in months).

“Critical age” at implantation for adequate emotion development

According to age at implantation children were so distributed into the 5 subgroups:

- 17 children implanted in 12 months group (mean age at assessment 6.435 years, sd 1984);

- 18 children implanted in 13e18 months group, (mean age at assessment 7.097 years, sd 1368);
- 13 children implanted in 19e24 months group, (mean age at assessment 8.346 years, sd 1781);
- 14 children implanted in 25e36 months group, (mean age at assessment 9.586 years, sd 2308);
- 10 children implanted in 37 months group, (mean age at assessment 10.187 years, sd 1271).

The Fig. X shows the mean z-scores of TEC according to age at CI defined into classes. Z-scores were significantly lower in the last three groups (19-24 months, 25-36 months, ≤37 months), indicating that subjects implanted within 18 months (12 months, 13-18 months) had better emotional comprehension skills ($p < 0.05$ by LSD post-hoc analysis from ANOVA). In ≤12 months group, all the 17 recipients performed within the TEC normal range; in 13-18 months group only 1/18 showed performance below the average. In the last three groups the number of children with poor emotional comprehension skills increased: 4/13 children in 19-24 months group (30%), 6/14 children in 25-36 months group (42.8%) and 4/10 children in 37 months group (40%).

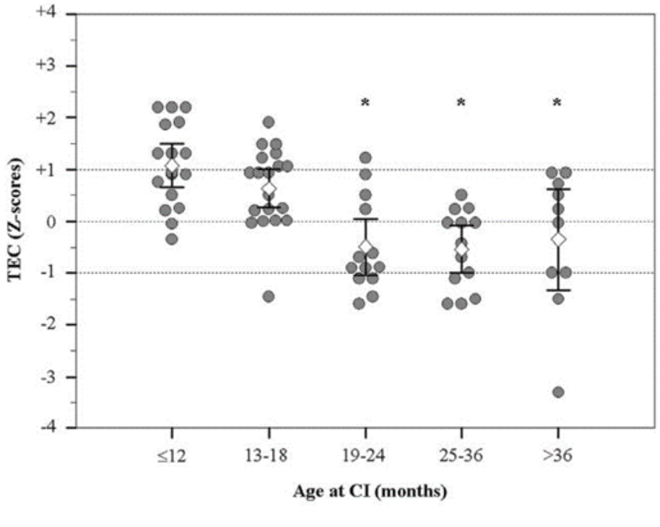


Fig. 10 TEC z-scores, according age at implantation; diamonds represent mean values; bars represent 95% confidence intervals; grey dotted lines represent the normal range in standardTEC z-scores, *$p < 0.05$ comparedwith the first class (<12 months) by LSD post-hoc test from ANOVA.

Furthermore, although children of ≤12 months and 13-18 months groups were younger and were using the device for a shorter time, their performances were the same or higher if compared to the other three groups, showing a development of these skills in time with their chronological age and in some cases also to a level above the average (7 children in 12 months group e 7 others in 13-18 months group performed $z > 1$ vs 1 in 19-24 months group and nobody in 25-36 months and ≤37 months groups).

Discussion

Emotion comprehension skills

The main aim of this study was to describe emotion comprehension skills, ranging from simple facial expression identification to morality awareness, in a wide cohort of deaf children using cochlear implants with an average device use of 6.1 years. To do this, the authors used the model described by Pons et al. (2004), where all these emotional components are explored, in order to obtain a comprehensive assessment of emotion comprehension development in children. In this model emotion comprehension development is thought to follow three developmental stages, external, mental, and reflective.

Our results highlighted that 79.17% of CI children in our sample mastered a degree of emotional comprehension skills that could be considered within the normal range in relation to their chrono-logical age (z-scores > -1 according to the TEC Handbook) (Albanese & Molina, 2008). Furthermore, in the younger age group, besides reaching the expected Pons' Stages 1, 2 or 3 according their chronological age, they also showed skills somewhat higher compared to NH peers. The remaining 20.83% performed below the normal range, with some children (4.2%) failing items of both Stages 2 (mental) and 3 (reflective) and the remaining showing difficulties only in the last and more difficult Stage 3 (16.63%).

All CI children were skilled in those aspects that are usually developed by NH children within 4-5 years of age (Stage 1, External): they correctly identified emotion by facial expressions, understood external causes of interior emotional status and were able to associate different desires to causes of different emotions. These basic competencies are those originally investigated by Wiefferink et al. (2013) and Ziv et al. (2013). As a matter of fact, our data confirm those of Ziv et al. (2013) who found no significant differences between 23 NH and 20 oral speaking implanted children. On the contrary, they are apparently in conflict with Wiefferink's data (2013), reporting significantly lower proficiency in 57 CI children when compared to 52 NH peers. Age at testing and auditory experience could be the factors influencing such findings, just as occurs in speech perception and communication (Tajudeen et al., 2010; Robinson et al., 2012; Geers et al., 2011). Wiefferink's study (2013) analyzed only children aged 2.5-5 years, and his CI sample included participants ranging from very short to medium follow-up of device use (range 1-44 months), while our sample included a wider range of ages (4-12 years) with medium-long follow-up (range 3.7-10.6 years). It could be assumed that at the time of implantation deaf children have a delay in the basic skills of identification of emotion and of understanding emotion from contextual situations, which are developed with increased auditory experience. CI subjects belonging to our study group mastered the Stage 2 (Mental) with outcomes that were within the normal value of the Italian Normative sample (Albanese & Molina, 2008): they were able to understand that different desires can cause different emotions, to differentiate between apparent and real emotions and to hide their own emotions or to identify dissimulation in others.

A different picture comes into play when considering more complex competencies belonging to the Stage 3 (Reflective), which is usually mastered by 8 years of age in NH children and that were a challenge for some implanted subjects. 16.63% of children, although having a chronological age between 8 and 12 years and a longer period of device use, still experienced a delay in this stage, showing difficulties in understanding multiple/ambivalent emotional responses to a given situation, in using psychological emotion regulation strategies, and in understanding moral rules.

There is basically a lack of research data concerning the complex task of understanding mixed emotions in NH children and no data at all in deaf children. Based on what we know about NH children, it might be speculated that, in some cases, deaf children facing mixed emotions tend to oversimplify them. As most emotion-evoking events could contain more than one element causing an emotional reaction (Aaker, Drolet & Griffin, 2008), understanding multiple emotions requires an increasingly sophisticated ability to differentiate between emotions and identify their elicitors, above all when emotions have the same valence. To perform these attributions in more complex emotion-eliciting conditions, there is a need for higher cognitive processing: appraisal and evaluation, comparison, categorization, inference, judgment/decision, belief, memory anticipation, and implicit cognition (implicit perception, implicit learning, and implicit memory) (Zajdel et al., 2013).. All these skills, that fall under the domain of executive functions, were found to be at risk of delay in deaf CI children (Conway, Pisoni, & Kronenberger, 2009; Kronenberger et al., 2013).

Hence, it might be assumed that owing to probable difficulties in executive functions tasks, the CI children failing the Stage 3 could engage in a more superficial processing of the emotion-evoking events. Once they have identified the most salient valence domain, they could inhibit the analysis, failing to perceive the concurrent existence of multiple emotions.

As far as regulation strategies are concerned, Rieffe (2012) reported that signing deaf children of hearing parents showed impaired capacities. The author reported that these children had difficulties in differentiating between their own emotion within the negative spectrum, in using avoidant strategies to cope with negative emotion, and in showing a strong preference for approaching the situation at hand, suggesting a more generic evaluation of situational difficulties. Similar conclusions were re-ported in the Wiefferink et al. paper (2013), where children were found to express negative emotions more often and more intensely, and to be less able to divert their attention from situations that generate discomfort, uneasiness, frustration, or grief. The same study group was the subject of a further analysis of the development of moral emotion expression (Ketelaar et al., 2015). The authors showed significant differences between NH controls and CI children, who expressed shame, guilt, and pride to a lesser extent and used a more reduced emotion lexicon. Once again, the short CI follow-up that characterized these two studies might explain differences with our findings. As for the more basic aspects of emotion comprehension, longer CI experience might have helped most children in our study group in attaining the normal range even in more complex emotional skills. For those who did not we might call upon various audiological and personal factors.

Factors affecting emotion comprehension

The second aim of this study was to identify personal and audiological factors affecting performances: presence of siblings and order of birth, gender, type of rehabilitation, oral language skills, severity of deafness, side of implant, type of implant and speech coding strategy, age at implantation, listening mode, and disyllabic word recognition.

The first-born and the only children scored better in items that define the achievement of the reflective stage. The effect of birth order in influencing child's development has been studied in NH children by Nelson et al. (2006), who explained that with increasing numbers of siblings there is a negative effect on the development of the youngest ones, likely due to the increased demands on parents' time and resources. The same effect was observed by Sarant and Garrard (2014) in deaf bilateral CI users and was associated with a significant reduction of performance in lexical comprehension, by approximately 5%, for each older sibling. Deaf children are at risk of missing out on the enriching aspects of “overhearing” conversations due to their auditory limitations and need more frequent and planned opportunities for receiving directed talk by their parents to learn language in optimal conditions (Cole & Flexer, 2007).

Regarding gender differences, it has been reported that NH girls aged 3-5 years seem more skilled than boys in understanding complex emotions (Bosacki & Moore, 2004). On the contrary, in our sample gender did not correlate with emotion understanding. Differences from previous studies (Bosacki & Moore, 2004) might be related to the age of the children included in our sample, which was on average greater than in Bosacki and Moore's study: it is possible that differences recorded in younger children catch up with age as has been already found for language development (Bosacki & Moore, 2004)

Type of rehabilitation was not related to performance mainly because the sample was not balanced according to this variable, with a broad group of children attending Auditory Verbal therapy (50 subjects) or Aural-Oral Programs (17 subjects) and only 5 rehabilitated with oral language supported by ISL. Oral approaches give more emphasis to listening and oral language development (Geers et al., 2011; Saarni, 1999; Ely, 2005; Estabrooks et al., 2006) and together with quality of parents' interaction (Landry, Smith &Swank, 2006) are important factors in emotion development. In particular, AVT is an approach that requires a deep involvement of parents that are educated and supported in the optimal way to communicate with their deaf children (Estabrooks et al., 2006) and therapeutic objectives, which are agreed by speech therapists and parents, cover all the areas of development, from cognition to communication and pragmatics, with great attention to socio-affective development. Also, this could be another reason for differences from Wiefferink et al.'s study (2013), where the sample composition included 53% of CI children that used oral þ signed language, 18% signers, and only 29% of subjects using exclusively oral language.

Oral language skills, lexical production in particular, were significantly related to the reflexive stage of emotion comprehension. These findings, similar to those observed in NH (Bosacki & Moore, 2004)] and pre-school CI children (Wiefferink et al., 2013), might be interpreted on the basis of the role played by language processes in interpreting and giving expression to both internal and external phenomena (Tangney, 2012). The development of language and emotion concepts is considered interdependent as both have their origins in social interactions with more skilled partners, during parent-child conversations, or free play with peers (Gergen, 2001). The richness of emotional language plays a large role in the development of children's conceptualization of emotions (Saarni, 1999) and provides them with a powerful tool for their comprehension and regulation (Kopp, 1989).

Regarding the audiological aspects, no correlations with TEC scores were found for severity of deafness, side of implant, type of implant, and speech coding strategy. A trend was found for better results in children with binaural listening, although the correlation was not significant (p = 0.062 and p = 0.053). Most recent studies highlight the benefits of bilateral cochlear implantation or bimodal stimulation in listening in everyday life situations characterized by frequent background noise, hearing at a distance, and group conversations (Zeitler et al., 2008, Lopez-Torrijo, Mengual-Andres & Estelles, 2015). Thanks to binaural redundancy, summation, and the head-shadow effect (Hughes & Galvin, 2013) listening effort is reduced, giving more opportunities to make connections between emotion expression and the connected contexts in everyday experiences, just as happens for language learning.

Disyllabic word recognition and age at implantation were strongly correlated with performance, and together with linguistic competencies, were responsible for 53% of the variance.

Auditory skills are universally considered as being predictive of better development of communication and language in deaf CI children. Higher performance in speech perception tasks is positively related with novel word learning ability, and consequently, with better language development (Davidson, Geers, & Nicholas, 2014), that is in turn strictly linked with emotional development.

Analogue considerations can be done for the influence exerted by the age at intervention on emotion comprehension.

Thanks to early intervention, the gap between auditory and chronological age is reduced, improving the quality and quantity of daily verbal exchanges with their caregivers, in a developmental phase in which they can take maximum advantage of cerebral plasticity (Fulcher et al., 2012; Nicholas & Geers, 2013; Kirk et al., 2002; Dettman et al. 2007; Guo, McGregor & Spencer, 2015). When implantation is performed in the first year of age, children's ability to build associations for the sound patterns of words and their references is more efficient and similar to that of NH children (Houston et al., 2012) and most children can reach adequate language skills by 4-5 years of age (Fulcher et al., 2012; Nicholas & Geers, 2013), an age at which better receptive and expressive language abilities have been shown to be strongly correlated to higher emotional skills in NH children (de Rosnay & Harris, 2002).

Good auditory skills are also related to better perception of indexical information, that is, the part of the speech signal which is responsible for providing information on voice characteristics of the speaker (i.e., age, gender, dialect, speaking rate) and on intentional facets of the message, including emotional moods, which, together with acoustic cues of phonemes and syllables, are crucial for conveying meanings in social spoken communication (Geers & Nicholas, 2013). Recognition of emotion in voice and speech seems to be a challenge for CI children (Most & Aviner, 2009; Hopyan-Misakyan et al., 2009; Nakata, Trehub, & Kanda, 2012), even though in children implanted at a younger age (< 12 months), better performance was seen in discrimination of the nuances of both speaker identity and emotion, two skills which are highly associated with their well-developed social competence (Geers & Nicholas, 2013).

Finally, early intervention could have a positive impact on the quality of parent-child interactions (Fagan, Bergeson & Morris, 2014), that has been suggested as an important predictor of emotional development in children because sensitive and structuring parents are more likely to match their teaching effort to their child's focus of attention, interests, and thoughts, guaranteeing the contingency needed to allow the right connection between an emotional state, its referent or its cause. Parents' expressiveness and reaction to children's emotions, and parent-child discussion by means of a rich use of emotional vocabulary strongly influence children's ability to manage, understand, and display emotions in a social context (Eisenberg, 2007; Brownell et al., 2013).

Deafness often modifies the quality of communication between NH parents and the deaf child, with consequent alteration of their "intuitive parenting skills," which consist of self-confidence in child rearing and in child oriented parental behaviour (Cole & Flexer, 2007; Luterman & Kurtzer-White, 1999; Reichmuth et al., 2013). Hence, unaware parents are at risk to be more intrusive, directive, and less flexible during interactions (Cole & Flexer, 2007; Spencer & Meadow-Orlans, 1996; Harrigan &Nikolopoulos, 2002). Thus reciprocity in relationships is negatively affected (Harrigan &Nikolopoulos, 2002; Spencer, 2004) and the establishment of joint attention and tuned communication is less successful (Nowakowski, Tasker & Schmidt, 2009).

Modification of these behavioural traits has been demonstrated by recent studies on patterns of interaction in deaf children implanted below two years of age and their hearing mothers, with positive increases in responsiveness (Vanormelingen, Maeyer &Gillis, 2015), improvement in interactional synchrony, and in maternal skills which adapt language complexity to the child's communication level. Changes can already be seen in the first 7 months after CI activation (Fagan, Bergeson & Morris, 2014).

The improvements in responsiveness promote tuned and embellished interactions that can help deaf children to gain shared experiences with their parents, to develop a better Theory of Mind (ToM) (Sundqvist et al., 2014) and to improve the ability to make inferences (Nicastri et al., 2014), all aspects involved in emotion comprehension (Landry, Smith & Swank, 2006; DesJardin & Eisenberg, 2007; Seidenfeld et al., 2014).

Critical age

The third aim of the study was to identify, if present, a critical age for emotional development in CI children.

Children that received their CI prior to 18 months of age performed significantly well in all emotion comprehension tasks. This age seems to be a sort of divide, that discriminates levels of matured skills: children in groups < 12 months and 13-18 months not only scored perfectly in the average range when compared to the NH test sample, but showed z-scores equal or over 1 in the majority of subjects; in children implanted later, different subjects did not reach normal scores, and even when they did, the z-scores were often negative.

The age limit of 18 months at implantation, in order to achieve a complete emotion comprehension maturation, seems apparently higher than the age considered as a good compromise for communication and language skills development (12 months of age) (Nicholas & Geers, 2013).

This finding could be explained considering the fundamental role that the right hemisphere plays in emotion development during the first two years (Borod et al., 1998; Chiron et al., 1997).

After birth and during the first year of life, the first emotional experiences (attachment bond with the primary caregivers) are largely influenced by progressive development of motor and sensory systems and are mainly mediated by a non-verbal lexicon (facial expressions, gestures, vocal tone, and prosody), relying on the activity of the right hemisphere (orbito-frontal system) (Chiron et al., 1997). Oral language starts to assume a more important role after the second year of age, when dominance progressively becomes complementary between the two hemispheres (Abbassi et al., 2011).

It can be speculated that when CI is performed before the age of 18 months, children that could have taken imprinting from the "non-verbal affect lexicon", will soon be able to master their ability of emotion comprehension through language development. On the contrary when implantation is performed after 18 months of age, children reach the end of the second year of age, with a short hearing experience or no experience at all, and their listening and language skills are still significantly delayed, taking experiences mainly from visual and tactile sensory modes, losing the important role of language in this age (Abbassi et al., 2011).

Also, the behavior of parents in these late implanted children is at greater risk of negative involution, since when the gap between chronological and linguistic age increases, parents have a less clear idea of the right way that they should interact and communicate with their children (Cole & Flexer, 2007). Their attention is focused on language gap recovery and they might experience difficulties in regulating their behavior and talking about abstract concepts such as emotions with their CI children (Ketelaar et al., 2015). On the other hand, it is well known how the level of language skills are likely to affect interactions between children and their parents because: (1) language facilitates children's regulation of attention, emotion, and behavior, and (2) language skills facilitate communication with parents, enabling better interactions and reduced levels of family stress (Quittner et al., 2013). High levels of stress in parents are associated with poor social and emotional development as well as increased behavioral problems (CrnicLow, 2002). These levels are in turn linked to the parents' perception of their child's progress: as language skills improve, parenting stress decreases and their ability to regulate children's behavior increases (Petersen et al., 2013). So the slower rate of development perceived by parents of children implanted after 18 months of age could maintain higher levels of stress for longer periods when compared with parents of children implanted earlier (Sarant & Garrard, 2014).

Outcomes from the present study should be interpreted with caution and represent a preliminary step towards comprehension of the relationship between age at intervention, the time of implant use, and other factors that can influence development of more advanced emotional skills in children with CI. The overall study sample was wide enough, but the number of children in each age group was small, so it could be affected by the particular characteristics of the individual child, although statistical analysis was specifically chosen to reduce most of this selection bias. Also, as the upper age limit for the TEC was 11 years of age, older children had to be excluded thus losing possible insights from them. Although TEC evaluates different aspects of the development of emotional comprehension, it leaves out expression of emotion and the ability to make use of regulation strategies, that are both fundamental to social functioning and are seen to be at risk in the CI population (Wiefferink et al., 2013). Further significant aspects that might influence emotional development were left out of our study, such as evaluation of the quality of parent-child interactions in terms of responsiveness and sensitivity (Montague & Walker-Andrews, 2002; Boiger & Mesquite, 2012), emotional language used in everyday interactions (Ketelaar et al., 2015), and level of development of the Theory of Mind (Ziv, Most & Cohen, 2013). Finally, this study offers a picture of this group of children in a given moment, whilst no longitudinal data was collected to analyse the characteristics of the maturation process.

3.2 Cognitive valence of an early auditory scaffolding

The prevalence of congenital hearing loss in infancy is nearly 1,5 cases per 1000 live births and the prevalence increases up to 6 years of age as a result of a late onset, meningitis, or a not timely diagnosis. Approximately 30% of deaf children have an additional disability, most commonly, a cognitive impairment (Van Naarden, Decouflé & Caldwell, 1999).

Congenital hearing loss can adversely affect the development of adequate language, oral communication, cognition, restricting learning and educational progress with potential far-reaching, lifelong consequences (Kral & O'Donoghue, 2010).

It is known that in case of severe profound hearing loss, an early and timely cochlear implantation can alleviate the deficits in the auditory system and promote cortical maturation. As a matter of fact, a considerable number of deaf children who receive an early cochlear implant reach normal cognitive levels and good language skills, with developmental trajectories similar to their hearing peers. However, a great variability of results in auditory, linguistic and academic outcomes still exists. This variability depends on complex interactions of variables, including age at onset of hearing loss, degree of hearing impairment, time of sensory deprivation, adequacy of restoring interventions, type and quality of intervention programs, social, parental and psychological factors.

After birth, cortical circuits undergo quite profound development, through synaptogenic and myelin maturation processes. In the first three- four years of life there is a peak in this synaptic count, with high plasticity of neuronal connections, profoundly susceptible to environmental experience. Stimulation of the auditory system during periods of maximal receptiveness (sensitive periods) is central to its normal development. Brain plasticity at age of cochlear implant is a factor significantly affecting neurodevelopmental outcomes: complex auditory functions and speech perception cannot be comprehensively established when hearing restoration is made late in life, since some aberrant developmental steps in synaptic counts, plasticity, and network properties have taken place without hearing (Kral & O'Donogue, 2010). An earlier intervention, exploiting juvenile plasticity, may prevent or reduce risk of dysfunction in brain maturation, increasing probability of an early and rich auditory and linguistic experience.

The importance of an early implantation relies firstly on the concept of auditory plasticity and sensitive periods. In NH children the synaptic density is highest at the age between two and four years. Density of dendritic trees reflects circuit changes and the increased power of cortical networks. After four years of age the synaptic counts decrease with elimination of not-used synapses: this phenomenon reflects the brain's need to specialize its functions for the prevailing demands. This specialization is accompanied by changes in synaptic potentials that have a longer duration in young subjects with a subsequent higher synaptic plasticity. On the other hand, in adulthood, synaptic potentials are shorter, leading to lower plasticity.

When a severe profound hearing impairment is restored late in life, auditory functions and speech perception cannot be comprehensively established because stimulation of the auditory system during sensitive periods of higher receptiveness is central to its normal development. On the contrary aberrant developmental steps in synaptic counts, plasticity, and network properties have taken place without hearing (Kral et al., 2006; Sharma et al., 2007). Delaying implantation beyond this temporal window can adversely affect brain adaptability, development of spoken language, communication skills with transversal far-reaching and lifelong consequences. As a matter of fact, an early auditory deprivation may have distal and detrimental effects on several neuropsychological and cognitive domains beyond language, affecting appropriate neural multimodal interactions that are crucial for a normal brain maturation

In normal hearing (NH) conditions, children gradually increase their ability to distinguish sounds, learning to abstract and categorizing them as auditory objects distinct from background noise. The place where these "auditory objects" take form is the cerebral cortex, especially the auditory cortex, made up of several functionally and histologically distinct Brodmann's areas. These interconnected areas are tightly linked each other, with lower-order areas activating higher-order ones (bottom-up interactions), and higher-order areas modulating those below (top-down interactions) (Kral & O’Donoghue, 2010). All these areas together constitute a one functional unit.

It is known that at birth the brain is the least differentiated of all organs. Phylogenetically, the expansion of human neocortex and higher brain specialization over time shaped the neural space for more and more complex communication skills; but brain anatomy and functionality need to be "enabled" to perform certain functions (Liotti, 2005). As Siegel (1999) correctly pointed out, a healthy neural and psychic development hinges on the possibility of receiving sensitive and tuned responses from its care environment. In fact, brain development largely depends on the encounter between genetically determined maturation programs and the richness and quality of the interpersonal and environmental experiences in which the child is immersed (Siegel, 1999). At a neuronal level, these experiences are impressed on the mind through neuronal activations that establish synaptic connections, determining their structure and functioning. Synchronized excited neurons tend to create connections between them, forming the neural networks of brain architecture.

With regard to this theme, the perspective proposed by Kral, Kroenenberger, Pisoni and O'Donoghue (2016) is to consider congenital deafness as a connectoma disease: that is, as an anomalous bias in the individual wiring and coupling pattern of the brain.

The brain develops through a dynamic and complex self-organizing process that includes a sequence of neurogenic events, including neurogenesis, neural migration, development neuronal connectivity and generation of central pathways. Development of the human connectome - the network map of neuronal connections comprising the nervous system (Petersen & Sporns, 2015) -is strongly linked to interaction between neural activity and environmental sensory experiences (Kral & O'Donoghue, 2010; Hübener &Bonhoeffer, 2014). This network of connections can be thought as a neural architecture constantly changing over time through "the four R's" activities: re-fixing; reconnecting; reforming and regenerating (Seung, 2012). During development, depending on different experiences and environmental stimulations, neural networks adapt or "re-fix" their connections by reinforcing or weakening them; neurons reconnect each other through creation and elimination of synapses; new circuits are reformed by growing of new ramification; new neurons are created and other delete. Hence, connectome is unique and different in different individuals since highly susceptible to experiences and external stimulations that are different for each one (Seung, 2012). Early auditory deprivation may compromise the synaptogenic process: congenital HL impacts on functional properties of the auditory system, affecting cortical development and connections between cortical areas (Kral &

O'Donogue, 2010). It also may interfere with central functioning which includes aspects such as cochleotopic representation, auditory space representation and cortico-cortical interactions. The auditory subcomponent of the human connectome (Figure 3.1) may include the substrate for procedural memory (connected with basal ganglia and cerebellum), also playing a role for declarative memory and spatial orientation (in connection with the entorhinal cortex and hippocampus), fear memory (in connection with amygdala), and attention.

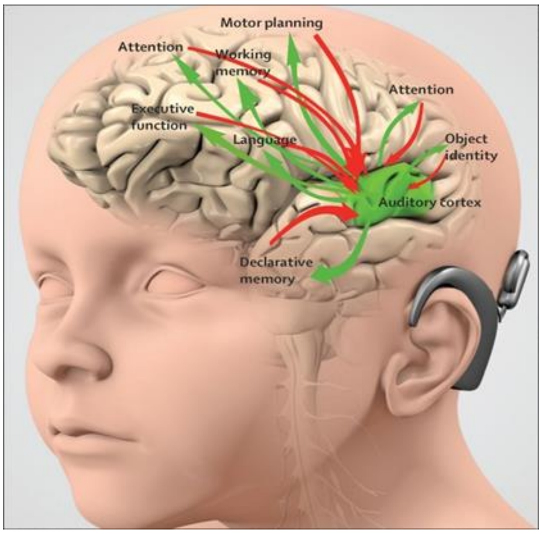


Figure 3.1: Auditory component of the human brain's connectome

Illustration of interactions of the human auditory cortex with higher order areas involved in cognitive functions. Locations of the functions on the brain are schematic. Bottom-up connections are shown in green, top-down in red (Kral et al., 2016)

A poor hearing experience in the first years of life may have widespread effects on brain development, affecting neural information processing and brain connectivity, not only in the auditory system, but also between different sensory systems: it may result in a sensory system reorganization, in stronger coupling to the remaining sensory systems or in reorganized interactions with other sensory systems, also affecting higher neurocognitive processes in a detrimental way (Kral et al., 2016).

Therefore, a profound hearing loss early in life may limit development of these skills, with a cascade of effects on perceptual and cognitive abilities, also beyond the processing of acoustic signals. Finally, losing sense of hearing may result in a reorganization of cortical functions (Bavelier, Dye & Hauser, 2006).

As a consequence, factors explaining the individual differences in clinical results after cochlear implant will not be limited to the auditory system itself: they could vary from the effects at the cellular level to those at the social level and reveal themselves in complex cognitive functions. For over 20 years, the “embodied cognition” theories have been emphasizing the close coupling of cognition, physical body and sensory systems: in other words, high cognitive processes, such as learning and memory, are strictly rooted in sensory surfaces, able to provide environmental sensory informations (Shneegans & Shoner, 2008).

Since the brain is an integrated self-organizing system, it appears clear that sensory deprivation should be thought as not fully independent from the rest of neurocognition: given the typical temporal and sequential quality of sound, it is possible to hypothesize that an early and rich auditory experience provides a sort of supporting framework, an “auditory scaffolding”, for the development of cognitive skills related to sequential processing (Conway, Pisoni & Kronenberger, 2009).

According to this hypothesis (Conway et al., 2009), the auditory experience would provide the primary gateway and the basic support to understand temporal and sequential events and for the development of spatial and temporal sequencing skills. A child who has continuous and repeated experiences with the perception and production of sounds gradually learns to codify and manipulate information sequentially. An early hearing deprivation may deplete cognitive abilities related to learning, recalling, and creating sequential informations, also affecting non-auditory functions related to time and serial order, including recalling task (Marschark, 2006) and executive functions.

A study from Conway found that CI 5-10 years old children performed worse than NH peers in motor sequencing tasks, showing atypical motor and visual sequence learning. Nevertheless, these CI children resulted as not impaired on several other non-sequencing tasks, such as visual-spatial memory and tactile perception (Conway et al., 2011).

As shown above, a poor hearing experience in the first years of life may have widespread effects on brain development. Electrophysiological studies showed a decreased maturation in left fronto-temporal and bilateral frontal areas (Wolff & Thatcher, 1990: in Conway et al., 2009) and a reduction of auditory-frontal connectivity (Emmorey et al., 2003), altering the neural organization of the prefrontal cortex.

A delayed maturation of prefrontal cortex may deplete not only motor and cognitive sequencing skills of deaf children: another crucial aspect to consider is the possible alteration of Executive Functions (EFs) that are a set of variably defined cognitive abilities.

As start point, we can define EFs as a group of top-down cognitive skills guiding planification, concentration, attention and inhibition of impulses. In a quite simplified model, EFs are a leading actions mechanism, based on inhibition of not relevant informations and upholding and mental manipulation of relevant ones. The EFs' domain was defined as an "umbrella term" (Lezak, 1995) to indicate multiple, interrelated and transversal cognitive abilities, considered as a family of self-directed and self-regulatory processes used in order to achieve a goal (Beer et al., 2014). At a neurobiological level, the executive control processes involve several brain circuitries: neuroimaging studies have shown that thalamus, basal ganglia and prefrontal cortex are mainly involved (Zuddas, Usala, & Masi, 2010). Thus, the EFs evolve along with development of prefrontal cortex, whose area covers up nearly 30% of total human cortex with a myelinization process running through adolescence and adulthood too (Valeri, Stievano, Ferretti, Mariani, & Pieretti, 2015), involving a lot of neural cortical and subcortical pathways (Steinberg, 2010).

A lot of definitions and theoretical models explaining EFs and their relations with cognitive, psychological, and linguistic development exist. It is beyond the purpose of the present work to delve deeply into this topic. Nevertheless, for the purpose of the present study, a brief summary of them may include three main theoretical frameworks: the "unitary models" (Baddeley, 1986; Norman and Shallice, 1986), the "sequential models" (Zelazo et al., 1997; Burgees et al., 2000), and the "fractionated-integrated models" (Miyake et al., 2000; Anderson & Reidy, 2012; Diamond, 2013).

Traditionally, in the unitary models (Baddeley, 1986; Norman and Shallice, 1986), the EFs are conceptualized as a unitary and general domain , assuming the existence of a supervisor central system exercising a strategic control over cognitive processes, selectively allocating attention and resources in order to achieve a task. Although there was some evidence to support the unitary EFs models, over time several researches were carried out, highlighting more complex and sophisticated functioning. The sequential models (Zelazo et al., 1997; Burgees et al., 2000) assume that different executive components come into play sequentially in solving problems or overcoming a complex task. According to the fractionated models (Miyake et al., 2000; Anderson & Reidy, 2012; Diamond, 2013), EFs should be considered as unrelated processes, with different developmental pathways.

An integrated model of EFs functioning has been offered by Miyake et al. (2000), assuming the existence of a partially dissociable common mechanism. This model has received broad scientific consensus suggesting that three basic dimensions of EFs exist: Working Memory — the ability to hold information in mind and use it; Inhibitory Control

— the ability to master thoughts and impulses so as to resist temptations, distractions, and habits, and to pause and think before acting- and Cognitive Flexibility — the capacity to switch gears and adjust to changing demands, priorities, or perspectives. These nuclear EFs may be the basis of other higher-order EF such as reasoning, problem solving and planning (Collins e Koechlin, 2012; Lunt et al.; 2012). Longitudinal studies have highlighted significant relationships between EF and: arithmetic and calculation ability (Blai e Razza, 2007; Espy et al., 2004), reading skills (Clark, Prior e Kinsella, 2002), verbal and non-verbal reasoning (Carlson, Moses &Breton, 2002; Van der Sluis, De Jong & Van der Leij, 2007), school performance (Biederman et al., 2004), social, moral and communication skills (Clark, Prior & Kinsella, 2002) and emotional regulation (Carlson e Wang, 2007; Simonds et al., 2007). Actually, their importance is strictly related to the efficiency in nearly every aspect of life, from infancy to adulthood and old age too. They provide critical supports for learning and development, allowing us to retain and work with information in our brains, focus our attention, filter distractions, and switch mental gears.

In next paragraph, recent findings from our research group on EFs and factors influencing their development in a group of preschool profoundly deaf children who received CI within two years of age will be presented.

3.3 Variables influencing executive functioning in preschool hearing -impaired children implanted withing 24 months of age: an observational cohort study

Abstract

Objectives: Executive Functions (EFs) are fundamental to nearly every aspect of life since they are needed to perform mental and physical actions or procedures, as well as to successfully fulfill complex operations. The present study was implemented to evaluate factors influencing their development in a homogeneous group of preschool orally educated profoundly deaf children of hearing parents, who received CI within two years of age.

Methods and materials: Twenty-five CI children, aged 3 to 6 years, were tested using the Battery for Assessment of Executive Functions (BAFE) to assess their flexibility, inhibition and non-verbal visuo-spatial working memory skills. The percentage of children performing in normal range was reported for each of the EF subtests. Mann-Whitney and

Kruskal-Wallis were performed to assess differences between gender, listening mode (mono and bilateral users) and degree of parents' education subgroups. The Spearman Rank Correlation Coefficient was calculated to investigate the relationship between EF scores audiological and linguistic variables.

Results: Percentages ranging from 76% to 92% of the children reached adequate EF scores at BAFE. Significant relations ($p<0.05$) were found between EFs and early intervention (age at diagnosis and CI), time of CI use, listening and linguistic skills. The Kuskal-Wallis test showed that children from families with parents who had had secondary school education and a university degree performed better at the response shifting flexibility task, inhibitory control and attention flexibility. Economic income correlated significantly with flexibility and inhibitory skills. Females performed better than males only in the attention flexibility task. No significant differences were found between mono and bilateral CI users for any of the EF tasks.

Conclusions: The present study is one of the first to focus attention specifically on the development of EFs in DHH children with CI in preschool age, providing an initial understanding of the characteristics of EFs at the age when these skills emerge. Functions such as working memory, shifting attention and inhibitory control are strongly linked to personal and social achievements, physical health and quality of life. Clinical practice must pay increasing attention to these aspects which are becoming the new emerging challenge of rehabilitation programs.

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Introduction

Executive Functions (EFs) are a set of top-down cognitive skills, which include response inhibition, self-control, interference control, working memory and cognitive flexibility. These mental processes provide critical support for learning and development, allowing us to retain and process information in our brain, focus our attention, filter distractions, and switch mental gears. Longitudinal studies have highlighted that EFs are at the core of school performance (St Clair-Thompson & Gathercole, 2006), emotional regulation (Carlson & Wang, 2007) and social, moral and communication skills (Clark, Prior & Kinsella, 2002).

Language plays a fundamental role in EF development as it allows children to share elements critical for the elaboration and expansion of mental images, facilitates the adaptation to environment requests and guarantees the inhibition of impulsive acting (Barkley, 1997). Good language knowledge is necessary to develop working memory, the executive function at the base of many cognitive operations, as it allows children to code external information to be then processed, stored, maintained, retrieved and transformed into phonological and lexical representations for use in a range of different processing tasks (Baddeley, 2007). Furthermore, the internal use of language, through self-reflection and self-questioning supports sustained and shifted attention, formation of rules and plans and control of behavior during problem solving activities (Remine, Care & Brown, 2008).

Given the relationship between language and EFs, deaf and hard-of-hearing (DHH) children represent an at-risk category for the development of these skills. Their limitations in receiving auditory information, accessing spoken language and using language for communicative purposes could affect their participation in daily communicative interactions from birth, negatively influencing neural organization and the development of domain-general neurocognitive skills that rely on auditory experiences, speech perception, and spoken language processing (Luria, 1973; Conway, Pisoni & Kronenberger, 2009; Pisoni, Conway, Kronenberger, Henning & Anaya, 2010; Kronenberger, Pisoni, Henning & Colson, 2013; Pisoni et al., 2008).

As a matter of fact, in the past twenty years, most of the studies on school age deaf population have highlighted a general delay in several areas of executive functioning, such as verbal working-memory (Kronenberger et al., 2013; Kronenberger, Beer, Castellanos, Pisoni &Miyamoto, 2014a; Kronenberger, Colson, Henning, & Pisoni, 2014b; Beer et al., 2014; Castellanos, Pisoni, Kronenberger & Beer, 2016; Davidson et al., 2019), visual sequence learning (Conway, Pisoni, Anaya, Karpicke & Henning, 2011), verbal rehearsal and fluency speed (Kronenberger et al., 2013; Pisoni & Cleary, 2003; Kronenberger et al., 2014a¸Castellanos et al., 2016), emotional and impulse regulation and inhibition-concentration skills (Kronenberger et al., 2013; Botting et al., 2017; Kronenberger et al., 2014a; Kronenberger et al., 2014b; Beer et al., 2014; Castellanos et al., 2016; Figueras, Edwards & Langdon, 2008; Barker et al., 2009; Beer, Kronenberger & Pisoni, 2011; Hintermair, 2013), sustained attention and attention shifting (Botting et al., 2017; Castellanos et al., 2016; Hintermair, 2013), sequential processing (Kronenberger et al., 2014b; Davidson et al., 2019); problem solving and planning abilities (Botting et al., 2017; Kronenberger et al., 2014b; Figueras et al., 2008; Hintermair, 2013; Marschark & Everhart, 1999).

Poor executive skills were detected both in DHH children using hearing aids (Botting et al., 2017; Figueras et al., 2008; Marschark & Everhart, 1999) and those with cochlear implants (Kronenberger et al., 2013; Pisoni & Cleary, 2003; Kronenberger et al., 2014a).

In preschool children with CI, EF skills were only studied by Beer et al. (2014). A sample of 24 DHH children who were implanted prior to age 3 and a control group of NH peers were directly assessed for short term working memory, inhibition and organization-integration skills. Furthermore, the Behavior Rating Inventory of Executive Function-BRIEF for parents (preschool version) was used to measure EF behaviors in everyday life (Gioia, Espy & Isquith, 2003).

Children's assessments showed significant differences between CI and NH in the domain of inhibition-concentration measures only, with a quarter of CI falling in the clinical range, against zero of NH peers.

For BRIEF, parents reported inhibitory control and working memory problems in almost 50% CI against the 15-30% of NH children. However, no statistically significant differences were reported in organization/planning. At the bivariate analysis, among all demographic and audiological factors, the duration of CI use was the only factor to correlate with fewer problems for planning and organization based on the parent-reported Plan/Organize scores of BRIEF. Language skills were positively associated with working memory and planning/organization skills, as measured through the BRIEF.

The preschool age represents the moment when core EFs are establishing, and the skills mastered in this period are strongly related to attentiveness, concentration, self-control, and ability to cope with stress and frustration during late childhood and adolescence (Moffitt et al., 2011) as well as physical health, financial well-being, and criminal outcomes in adulthood (Moffitt et al., 2011). Investigating such an early phase of competence development could be useful in developing adequate strategies of intervention aiming to reduce the long-term negative effect of early inadequate patterns of EF skills. Given the paucity of data on this phase of development, the present study was implemented to evaluate factors influencing the development of adequate EFs in a homogeneous group of preschool orally educated profoundly deaf children of hearing parents, who received CI within two years of age and have at least one year of auditory experience.

Material and methods

Participants

Children with congenital profound deafness (Pure Tone Average in the better ear ≥90 dB HL for 500–4000 Hz), aged 3 to 6 years at the time of enrollment, were included. In order to limit the number of variables that can be reasonably accounted for in the statistical analysis without introducing confounding interactions (Kahlert et al., 2017) and to eliminate variation in the confounders (Pourhoseingholi, Baghestani & Vahedi, 2012), the following inclusion criteria were introduced in subject selection: normal cognitive level, as assessed by Raven Colored Progressive Matrices (Raven, 1986; Raven,2003); absence of additional handicap and/or associated disorders verified by clinical history and neuropsychiatric evaluation; absence of pathologies/alterations that could impact the auditory outcomes of cochlear implant, such as cochlear and nerve malformations, auditory neuropathy, meningitis; Italian as primary household language; child oral education setting.

Economic income was defined on the basis of the Italian economic family status indicator index named ISEE (Indicatore della Situazione Economica Equivalente: Equivalent Economic Situation Index). The ISEE index allocates economic income brackets based on annual income, real estate assets, number of members of the family and city of residence (https://www.inps.it/nuovoportaleinps/). Based on this index three economic income brackets were defined: low, middle and high.

Study design

The present study was structured as a cohort longitudinal study and data were collected in two CI centers: Policlinico Umberto I Hospital, Rome and “Guglielmo da Saliceto” Hospital, Piacenza, Italy. The protocol was approved by the local ethics committees of the two hospitals. The recruited families gave written informed consent for the assessment of their child before commencing any study-related procedure. Protocol studies were approved by Institutional Review Board and was conducted according to the principles and rules laid down in the Declaration of Helsinki and its subsequent amendments.

Assessment

Auditory skills

Speech recognition was assessed using standard Italian phonetically balanced bi-syllabic words for pediatric populations (Cutugno, Prosser &Turrini, 2000). A 10 item list was preceded by a practice list. Items were administered in a soundproof room, via a loudspeaker placed at 1m distance from a table where the child was sitting next to a speech therapist. Stimuli were presented in quiet at 65 dB SPL 0° azimuth. Score was calculated as a percentage of words correctly repeated.

Language skills

Children were tested individually in a quiet room, by two trained speech therapists. Tests were conducted using spoken language as all of the children communicated orally. Two Italian Standardized Language tests were used to assess, respectively, lexical comprehension and lexical production and morphosyntactic comprehension.

Lexical comprehension and production were measured using specific subtests of “Test di Valutazione del Linguaggio -TVL” [Test for Evaluation of Language] for preschooler (Cianchetti & Sannio Fancello, 1997) designed for children ranging from 3 to 6 years of age. Children were presented real objects or pictures and were requested to point or name them. The raw score is represented by the number of correct responses, which are then transformed into normative weighted scores ranging from 0 to 10. A weighted score of < 3 is considered below the average (more than -1sd). Test retest reliability was respectively .91 and .96. for comprehension and production.

Morphosyntactic comprehension was assessed using “Prove di Valutazione della Comprensione Linguistica-PVCL” [Test for the Evaluation of Linguistic Comprehension] (Rustioni & Lancaster, 1994). PVCL is a test that investigates grammatical comprehension skills (for example reflexive, negative, passive, reversible, temporal, causal, conditional and adversative phrases). It is divided into protocols ordered by age groups (from 3.5 to 7 years). Each test stimulus is presented in a four-picture, multiple-choice format with lexical and grammatical distractors. For each item, the examiner reads a sentence that refers to one of four drawings. Children were asked to point to the drawing corresponding to the sentence presented by the examiner. A total raw score is calculated based on number of correct items identified, each of which has a different value according to its developmental complexity.

The raw score is then converted into a percentile. A percentile <25 is considered below average (more than -1sd). Test retest reliability was .93.

Executive Function skills assessment

The neuropsychological assessment of EFs was carried out using the Battery for Assessment of Executive Functions-BAFE (Valeri, Stievano, Ferretti, Mariani, & Pieretti, 2015), a battery specifically designed to assess nuclear EFs in preschool children, aged from 3 to 6. It is composed of four subtests, consisting of quick, easy and quite engaging activities for young children, with the aim of measuring three aspects of EF skills: Flexibility, Working Memory and Inhibition.

Flexibility (set-shifting) refers to the ability to shift, in a flexible manner, between different mental plans in order to reflect different situations or external requests. Usually subjects with reduced set-shifting when faced with a problem or new situation, show perseverant behaviors, mental rigidity and lack or reduction of flexibility. Flexibility is thought to be subdivided into two different skills: response shifting flexibility and attention flexibility. Response shifting flexibility is the ability to shift behavior from one mental set to another one that conflicts with the first (i.e. resolution of cognitive conflicts). Attention flexibility is the ability to focus attention on a mental set while resisting to interference. BAFE assesses these two aspects of flexibility, including two different subtests: Card Sort, for assessing response shifting flexibility, and Triplets of Circles Pattern making, for assessing attention flexibility. Both subtests evaluate flexibility with minimum language skills involvement.

For Card sort tasks the child is shown images that they have to categorize at first according to shape and then according to color. The images must be inserted in the appropriate containers. One point is given for each complete and correct answer (range 0-3): a complete answer is when the two criteria, shape and color, are present in the sample image (little blue bear and red house→ little red bear and blue house→1 point). The set-shifting ability is examined through the child's ability to switch from one categorization criterion to another during the test. One point is given for each correct response (range 0-3)

For Triplets of Circles Pattern making, the child is shown a series of colored circles printed on a strip of card, which are always repeated with the same sequence and they are asked to name the color of each circle (for example "blue, blue, red, blue, blue, red) for all of the sequences. The child is then requested to reproduce the overall sequence on a board using small plastic blue and red circles. One point is given for each correct triplet (range 0-6).

Inhibition refers to the ability to stop or delay impulsive/compelling responses, to self-control attention and emotions in order to achieve a behavioral adaptation goal. Moreover, it encompasses delay aversion or gratification skills and, more generally, the ability to wait. Usually, children with low inhibitory control show impulsivity and inefficient organization.

In BAFE, inhibitory skills are assessed by subtest Stroop-like day-night. It consists of a set of cards, presented to the child, where the moon or sun are alternatively depicted. The child is asked to say the word “day” when he is shown images with the moon, and the word “night” when he is shown ones with the sun. The child must inhibit automatic responses. One point is given for each correct answer (range 0-16).

Working memory refers to the ability to store and manage verbal and/or non-verbal information, in order to reflect complex cognitive tasks such as understanding, learning, reasoning. A deficit in working memory makes it harder to remember information, to plan actions in order to achieve a goal, to create mental representations and to make decisions. Working memory has a crucial role in selection, initiation, and termination of information-processing functions such as encoding, storing, and retrieving data. In BAFE, working memory is assessed in its visuospatial subdomain by subtest Spin the Pots.

The test is administered using a rotating tray with eight different colored cups. A red token is placed under each cup and a cloth is used to cover the game. The tray is rotated while it is covered. After it is rotated, the child is asked to lift the cloth and choose a cup in order to find a red token. The child has to recover all of the tokens, but without choosing the same cup more than once. The position of the cups changes each time because, after a red token is recovered, the cups are rotated on the supporting tray. The raw score is given by the number of attempts made to recover all tokens (range 8-16).

Raw scores for each FE subtest are converted into percentile ranking according the normative sample of the test. The BAFE manual references normal scores as ≥ 25 percentile.

The battery is standardized on a sample of 358 children (167 girls, 191 boys), balanced for gender and subdivided into six age-groups. BAFE shows a good reliability coefficient for subtest Card Sort and Spin the Pots (KR-20= 0.77), and excellent reliability coefficients for subtest Day and Night (KR-20=0.92) and subtest Triplets of circles (KR-20=0.94).

Statistical analysis

Statistical descriptive analysis is presented as median [min. and max.] for continuous variables. CI outcomes were compared with scale norms for the test batteries (which are based on nationally representative samples for typically developing, normal hearing children). The percentage of children performing in normal range was reported for each of the EF subtests. Consistently with the non-normal distribution of data the Mann-Whitney and Kruskal-Wallis tests were used to assess differences between gender, listening mode (mono and bilateral users) and degree of education subgroups. The Spearman Rank Correlation Coefficient was calculated to investigate the relationship between EF scores and demographic, audiological and linguistic variables. P-values less than 0.05 in either direction were considered as significant. Statistical analysis was carried out using a PC version of Statistical Package for Social Sciences 25.0 (SPSS, Chicago, IL, USA).

Results

Subjects

Twenty-five children (15 females, 10 males) met the inclusion criteria. They had a median age at assessment of 5 years (min 3.3-max 5.9 years). Table 1 shows their main demographic and clinical characteristics.

All children had a profound congenital sensorineural hearing loss caused by Connexin 26 mutation (11), ototoxicity (4) and unknown etiology (11). Median chronological age at diagnosis was 4 months (range 1-21). All children started a habilitation process within one month of diagnosis. Median age at implantation was 12 months (range 8-24). Sixty-four per cent of children were diagnosed within 6 months and implanted within 12 months of age. Of them, 50% received CI between 8 and 12 months of age. The median time of device use at assessment was 48 months (range 15-59). Fourteen recipients were implanted with Cochlear devices programmed with ACE strategy and 11 with Advanced Bionics devices programmed with Hi-Resolution 120 strategy. Seventeen children used bilateral CIs (7 simultaneous and 10 sequential), while 8 were unilateral CI users.

The median normalized score for non-verbal intelligence at CPM was 82° percentile (range 42-100). Concerning communication mode, all children attended oral rehabilitation programs and were completely immersed in an oral communication environment. All of them used oral spoken language and attended normal mainstream kindergarten with the presence of a support teacher, according to the normal legislative procedure of the Italian Ministry of Education.

Median bysillabic word recognition in quiet was 100 (range 70-100). Eighty-eight per cent of children had a maximum score in bysillabic word recognition.

TVL median weighted scores for lexical comprehension and production were respectively 7 (range 1-9) and 5 (range 1-9). Eighty-eight per cent of children fell into the normal range for lexical comprehension, and 76% showed normal scores for lexical production. The median percentile score for morpho-syntactic comprehension (Rustioni test) was 75°: 92% children fell in the normal range. Concerning parents' education, median value for school attendance was 13 years (range 8-18) equivalent to a high school level, and 40% of mothers and 36% of fathers had a university degree. Most of the sample was constituted of families with medium economic income (72%). Between the remaining families, 4 (16%) and 3 (12%) had respectively a lower or a higher economic income.

Executive Function domains

The median score in the Card Sort task, for response shifting flexibility, was 99 percentile (range 10-99), with 22 children (88% of recipients) showing adequate performance (≥25 percentile): of these, 18 (72%) obtained an optimal score (≥70 percentile). A similar trend was shown in the Triplets of Circles task, for attention flexibility: median score was 99 percentile (range 5-99), with 23 children (92%) reaching sufficient scores and 22 of these (88%) within optimal range performance.

Concerning the Stroop-like Day-Night, for Inhibitory Control, median score was 48 percentile (range 5-99), with 19 children (76% of recipients) performing in the range ≥25 percentile. Seven children (28%) showed high level performance.

Finally, median score for the Spin the Pots task, for visuo-spatial working memory was 43 percentile (range 5-99), with 20 children (80% of recipients) showing a normal range performance. Four children (16%) obtained optimal working memory score.

Children who were identified within 6 months of age and received CI within 12 months of age reached ≥25 percentile performance in 94% of cases for response shifting flexibility and inhibitory control and in 100% of cases for attention flexibility and visuo-spatial working memory skills. Children who received diagnosis and were implanted later showed more variable performance, with fully sufficient skills in 78% of cases for response shifting and attention flexibility tasks and only 44% for inhibitory control and visuo-spatial working memory.

Factors that affect EFs

Bivariate correlation was performed between EF outcomes and children's demographic (age at diagnosis, age at cochlear implantation, period of CI use, bi-monolateral CI, economic income and years of parents' schooling), audiological (bisyllabic word recognition in quiet) and linguistic variables (lexical comprehension and production and morphosyntactic comprehension).

Language skills correlated significantly with all the EF outcomes. Age at diagnosis and at CI showed negative correlation with inhibitory control and both response shifting and attention flexibility, while they seemed not to impact visuospatial working memory. Children with longer CI use showed better skills in flexibility tasks only. Listening skills were correlated with EFs, with the exception of visuospatial working memory.

The Mann Whitney test revealed no differences in performance due to gender with the exception of the attention flexibility task which was significantly better for females. No significant differences were found between mono and bilateral CI users for any of the EF tasks. Parents' level of education was analyzed according to the achievement of a junior, high school or university degree. The Kruskal-Wallis test showed that children from families with parents who had had secondary school education and a university degree (respectively 13 and 18 years of education) performed better at the response shifting flexibility task, inhibitory control and attention flexibility. Economic income correlated significantly with flexibility and inhibitory skills.

Discussion

The aim of the present study was to evaluate factors that influence EF skills in preschool congenital profoundly deaf children, who received CI within two years of age.

The first variables studied were early diagnosis and early cochlear implantation. The behavioral flexibility, attention flexibility and inhibition skills significantly correlated with both age at diagnosis and age at implantation: children who were diagnosed and implanted early had better skills and were more likely to perform within normal range. Our findings differ from those described by Beer et al. (2014) in preschool children and from other studies on school aged children (Pisoni & Cleary, 2003; Davidson et al., 2019; Kronenberger et al., 2014a; Kronenberger et al., 2014b), while are in agreement with data reported by Conway et al. (2011). Pisoni & Cleary (2003), Beer et al. (2014), Davidson et al. (2019) and Kronenberger et al. (2014a; 2014b) included in their analysis children with a different age range of onset of deafness (0-36 months), variable pre-implant PTA (ranging from about 70 dB to 120 dB) and ear congenital malformations. When these variables are not controlled in the analysis, the effect of early intervention could be lost due to good outcomes in children, who despite having a late CI, had a pre CI hearing experience (Geers, Nicholas, & Moog, 2007). Conversely poor outcomes are possible in children with early implantation and diseases that limit their benefits (Demir et al., 2019; Shearer & Hansen, 2019). As in the present study, Conway et al. (2011) only included children with bilateral profound hearing loss with no residual hearing prior to CI. A significant negative correlation was found between implicit sequence learning abilities and age at implantation: children with the least auditory deprivation and earlier CI showed better visual sequence learning outcomes.

In the present study, a significant percentage (64%) of children were diagnosed before 6 months and received cochlear implant within 12 months of age (50% between 8 and 10 months). All of these children reached normal performances (≥25° percentile) in Attention Flexibility and Visuo-spatial Working Memory. Ninety-four per cent of them also performed as expected for their chronological age in Behavioral Flexibility and Inhibition control. On the other hand, children diagnosed and implanted later showed more variable performance, with 56% of them scoring <25 percentile in Inhibition Control and in Visuo-spatial Working Memory and <22 percentile in both Behavioral and Attention flexibility. To our knowledge, this is a novel finding, as it underlines how receiving CI within 12 months of age influences EF skills when compared to children implanted later, in agreement with findings reported in the auditory and communicative domains (Dettman, Pinder, Briggs, Dowell & Leigh, 2007; Colletti, Mandalà, Zoccante, Shannon & Colletti, 2011; Leigh, Dettman, Dowell & Briggs, 2013). One possible explanation is that earlier access to sound is fundamental to activating attention-demanding systems (Conway et al., 2011) and also to fully develop some basic cognitive mechanisms, important for learning, such as sensory integration (Houston, Stewart, Moberly, Hollich & Miyamoto, 2012). In fact, the experience acquired in the early stages of life with sound and auditory patterns, which are complex signals and arranged in series, gives the child the opportunity to develop the bases for neurocognitive and executive functions such as: detection of patterns, sequential memory, sustained attention, cognitive flexibility, planning and problem solving (Conway et al., 2011; Kral, Kronenberger, Pisoni & O'Donoghue, 2016). Early identification and early implantation significantly reduce deprivation and give children the opportunity to learn from exposure to complex auditory stimuli in the period of maximum brain plasticity (Kuhl,2010).

Visuo-spatial working memory didn't correlate either with age at diagnosis nor with age at implantation. The children in the present study were all preschooler, with a maximum age of six, and still using a visuospatial code - not affected by auditory deprivation: this could explain the lack of correlation. Young children's visuo-spatial working memory seems to rely above all on the ability of the child to visually store perceptive characteristics of the materials they need to memorize (Pickering, 2001). They use information such as shape, orientation and detailed appearance (Pickering, 2001) but are not yet able to form a verbal mental model of the objects. This reliance on visual information is gradually replaced by the verbal rehearsal of visual and spatial cues of objects by age of seven (Fenner, Heathcote & Jerrams-Smith, 2000).

Speech perception was also associated with all of the EF abilities studied, with the exception of the domain of Inhibitory Control. Beer et al. (2014) didn't investigate the relationship between EFs and speech perception in their sample of preschool children, while some connection was found in the few studies that focused on this aspect in school children. The relationship seems to be closely linked to the type of EF domain being investigated (Pisoni et al., 2010; Pisoni & Cleary, 2003; Kronenberger et al., 2014b; Beer et al., 2011): direct or inverse correlations were found with verbal working memory, verbal rehearsal speed (Pisoni et al., 2010; Pisoni & Cleary, 2003; Kronenberger et al., 2014a) and behavior regulation (Beer et al., 2011), but not with spatial working memory and inhibition-concentration (Kronenberger et al., 2014a). A possible explanation could be the differing complexities of the speech perception tasks being performed and the degree of verbal demand of each EF task. For example, speech in noise tasks are more complex than speech in quiet ones as the first are regulated by a child's ability to focus attention on the speech signal, correctly encoding, storing, and reproducing words in the sentence while simultaneously inhibiting the distracting noise. Therefore, they are likely to be more effective in identifying relationships with various EF skills, such as inhibition-concentration (Beer et al., 2011).

Language skills correlated significantly with all the EF domains studied.

CI children with better language, as assessed through language tests, are more able to switch between verbal and perceptual domains, and between different sorting concepts within each of the domains (Remine et al., 2008; Figueras et al., 2008). Further, language helps children regulate behavior, concentrate and inhibit impulsive responses (Figueras et al., 2008). Internal and external language is used to reflect on events and tasks, and it allows for the decoupling of action from reality. Direct behaviour can therefore be guided by action plans, that are stored internally in the working memory, rather than by immediate external factors (Figueras et al., 2008). Poor language skills can therefore make it difficult to memorize the rules and the phases of a plan and can induce children to give more impulsive or hasty answers (Botting et al., 2017). Unfortunately, the lack of a NH control group did not allow to investigate the direction of the relation.

The data in the literature on DHH children are, instead, conflicting about the relationship between visuo-spatial working memory and language. Spatial, visual and language skills were positively correlated in the studies of Surowiecky (2002), Edwards & Anderson (2014), Ulanet et al. (2014); Figueras et al. (2008) and Jones et al. (2020). In particular, a longitudinal study (Jones et al., 2020) revealed a developmental path that suggests that visuospatial working memory does not develop optimally when the child's existing vocabulary is weak. In other studies, visuo-spatial WM was less dependent on the development of the first lexical organization and, therefore, more resistant to delays and early hearing disorders (Kronenberger et al., 2013; Kronenberger et al., 2014a; Kronenberger et al., 2014b; Castellanos et al., 2016; Lyxell et al., 2008). Visuospatial WM tasks may differ in the degree of sequential processing of stimuli. Despite the seemingly non-verbal design of the task, children could still make use of verbal mnemonic strategies such as labelling stimuli or naming aloud the position of a stimulus (Surowiecki et al., 2002). The visuospatial task we used in the present study may have benefited from these verbal strategies, and children with better language skills may have obtained higher scores through their use, thus helping to establish a positive correlation between language skills and visuospatial WM performance.

Gender seems to play a role only for the attention flexibility task: females appeared to have better skills in focusing attention on a mental set, resisting interference. Although there are no other studies on gender difference and EF performance in children with CI, studies focusing on children with NH have shown that girls would perform better in verbal working memory and attention, while boys would achieve better results in terms of spatial reasoning/working memory and cognitive flexibility (Overman, Bachevalier, Schuhmann & Ryan, 1996; Seidman, Valera & Makris, 2005; Memisevic & Biscevic, 2018).

The role of economic income has been poorly studied. In the present study group of preschool CI children, it was significantly correlated to flexibility and inhibitory abilities. This finding concurs with the outcomes described in school-aged DHH children by Mitchell

& Quitner (1996). On the contrary Beer et al. (2014) found no correlation between the studied EFs and these socio-demographic variables. Studies on NH children have shown that low economic income is associated with worse performance in inhibitory control tasks, working memory, executive attention as well as flexibility and planning (Lipina, Martelli, Vuelta & Colombo, 2005; Mezzacappa, 2004; Noble, McCandliss & Farah, 2007; Raver, Blair & Willoughby, 2013; Hackman, Gallop, Evans & Farah, 2015). The influence of economic income is already traceable in infants (Clearfield & Jedd, 2013) and pre-schoolers (Raver, Blair & Willoughby, 2013; Blair et al., 2011; Hughes, Ensor, Wilson &Graham, 2010; Noble, Norman & Farah, 2005), and this persists over time (Hackman et al., 2015). Given the initial premises and results described in this work and in the one by Mitchell & Quitner (1996), it would be important that future prospective studies investigate further the role of economic income in the FEs of children with CI.

Finally, in the present study it was found that parental education level influences the EF skills of children with CI: children of parents with a higher education level have been positively associated with flexibility of response change and inhibitory control. This new finding correlates with the linguistic results described by Geers, Nicholas and Sedey (2003) in a large population of CI children, subsequently confirmed by Cuda et al. (2014) in preschoolers. Parental education level has been described as a very important factor influencing NH children's cognitive development (Ganzach, 2000), and also predicting the correct development of EF performance (Ardila, Rosselli, Matute & Guajardo, 2005). Parents with a higher education level create a more stimulating environment from an intellectual point of view (Hoff, 2003), establishing the environmental conditions that favour the development of Efs (Hoff, Laursen & Tardif, 2002). They talk more, read more often to their children and use a richer vocabulary than parents with lower educational levels (Hoff-Ginsberg,1991). Children of parents with higher education levels tend to have a wider vocabulary, faster language development and better performance in cognitive tests, including EFs (Ardila et al., 2005).

Some constraints limit the generalization of current results. Despite being homogeneous, the sample was small and only limited statistical analysis could be performed. The EF assessment was performed solely through a direct test of the children and this limited the comparison with other studies that integrated parental behavior checklists to measure additional EF domains in daily life. Finally, the study assessed EF skills at a given time, preschool age, but longitudinal data are not yet available, leaving unanswered the question as to whether these skills are maintained at an older age, when basic competences mature and further and more complex skills will require a higher cognitive load (Kral et al., 2016).

Conclusion

The results of this study highlight that early intervention, language development, economic income level, gender and parental education all play a role in determining the development of EFs in preschool CI children. Most of early implanted children developed good EF skills. However, 6% of them showed difficulty in flexibility and inhibitory control. Evaluation of EF skills should therefore become a routine aspect of follow-up immediately after cochlear implantation, and intervention programs should include strategies and specific training to enhance and monitor these skills. A timely identification of developmental difficulties, if present, could allow professionals to implement an appropriate stimulation program for the child, therefore indirectly helping families to put into practice behaviors able to enhance EF competences. Further longitudinal studies would be useful to add information on the variables that influence the development of EFs and to improve the effectiveness of intervention strategies.

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Chapter 4. Hearing loss and cochlear implantation in elderly: a holistic perspective to quality of life

4.1 Aging, old age and hearing loss: overview and clinical manifestations

From a biological point of view, aging can be defined as a decline or a “de-tuning” (Rose et al., 2007; Rose, Flatt et al., 2012) of adaptation, caused by a time-progressive decline in intrinsic physiological function.

Aging is undeniably a biological process with its own dynamic, largely beyond human control (Gorman, 2000). Anyway, the large inter-individual and intra-individual variability makes it extremely difficult to define *a priori* and rigidly the age in which a person is “old”. In fact, in addition to the intertwining biological, cognitive, social, cultural variables that define everyone as unique and different, even in the same person the biological, psychological, cognitive and social age are often not coincident aspects. In aging, cognitive and psychological components, although interrelated and mutually influencing, cannot be superimposable. A cognitive well-functioning old person does not necessarily coincide with a psychological well-functioning old person.

The loss of interests, self-absorption, withdrawal in deformed memories of the past, isolation and loneliness are psychological experiences in many old people. A good cognition does not protect necessarily against maladaptive emotional states: variables that come into play refer to personality traits, individual history, social and family environment, specific life-stressors and personal resilience skills.

Conventionally, from a social and scientific point of view, the age of 65 years is said the beginning of “old age”, roughly corresponding to retirement ages; however, in recent years and especially in western societies, this threshold is moving forwards, increasingly shifting towards the age of seventy. It remains however an arbitrary threshold, more conventional than realistic, accounting more for chronological parameters than for processes embedded in aging.

Since the 1960s, theoretical approach to aging gradually evolved from a decline-oriented approach - focused on functions declining with age - towards a development-oriented approach, focused on functions that remain sane or possibly improved. The Life

Span Development Theory (LSDT; Baltes and Baltes, 1986; 1990; Baltes and Silverberger, 1994) has made a significant contribution towards this crucial theoretical shift: from development as a short-lived process characterized by a gradual global decay, to development as a non-linear life-time process, whose developmental trajectories may differ for a variety of biological, personal and social conditions. From this point of view, whatever developmental stage -from infancy to old age- everyone has developmental resources, his own potential to evolve that need to be optimized.

Researches across neurophysiology and neuropsychology strongly supported the LSDT approach, on the one hand refuting a "global decline" hypothesis, on the other, highlighting the impact of experience and environmental factors in brain and mental changes, pointing out for example a not always linear correlation between brain deterioration and residual cognitive skills (e.g., Salat, Kaye and Janowsky, 2002; Mitchell et al., 2009).

From this point of view, whatever developmental stage -from infancy to old age- everyone might have developmental resources, his own potential to evolve that need to be "optimized".

In recent years, there has been an increasing interest in the possibility of modifying these aspects through interventions – combining neuroscientific and psychological approaches to cognitive aging- able to exploit residual brain and cognitive plasticity. Following the theoretical approach of the Scaffolding Theory of Aging and Cognition (Park & Reuter-Lorenz, 2009; Reuter-Lorenz & Park, 2010), even in adult/old age, despite the physiological deterioration of brain and neural structures, there is a significant margin of cognitive plasticity through which the cortical structures respond to deteriorative changes by reorganizing alternative networks in order to preserve adequate cognitive functioning. This "neural scaffolding" -or support framework- would occur through the recruitment of alternative or complementary circuits (activation of counter-lateral homologous areas, increased frontal activation) to those in deterioration or functionally damaged. The mechanisms that can lead to the expansion and improvement of this neural scaffolding would lie in the neural response to external experiences, such as exposure to cognitive exercises and training, an increase in social and relational investments, or a renewed commitment to complex tasks.

The "Theoretical Perspective on Cognitive Aging" (Salthouse,1991;1994) pointed out that cognitive decline in aging, far from being a general deterioration, may result from a decline in speed of processing information, with a subsequent reduction of mental resources to process new stimuli. Processing speed is defined as the rate at which a person can understand and react to an information; the time he takes to do a mental task. It is considered one of the main factors explaining the differences that advancing age generates in cognition throughout the course of life. This decline has been attributed to changes in weight of the brain, in the dendritic structure, in fibers myelination and in a lower effectiveness of dopaminergic receptors that would lead to a "slowdown" in cognitive operations and to deteriorated performance (De Beni & Borella, 2015).

According to this global/macro approach to aging, deterioration of speed processing in elderly might be the basis of worsening in elderly performance, resulting in slower mental operations and problem solving, in less reasoning accuracy, in worsening of working memory and episodic memory (Salthouse, 1994; Salthouse, Berish and Miles, 2002). Salthouse's hypothesis partially takes up the previous multidimensional hypothesis of aging by Horn and Cattell (1967): Authors hypothesized that only fluid skills suffer a cognitive deterioration; on the contrary, crystallized functions (cultural and learned acquisitions) do not undergo deterioration. Hence, when cognitive tasks require processing of unfamiliar stimuli, fluid skills as reasoning, working memory and perceptual skills are involved, with depleted performance in elderly. In contrast, when cognitive tasks require involvement of automatic skills (cultural, verbal, numerical knowledge), performance shows a mild or poor deterioration (Jennings & Jacoby,1993; Noack, Lövdén, Schmiedeka & Lindenberger, 2009). Decline of all the components of intelligence would therefore occur later in life, when also the crystallized compensatory effect slows down or ceases to function.

4.1.1 Age-related hearing loss and cochlear implantation

When hearing loss occurs in elderly, its weight and repercussions on patients 'general functioning and wellbeing need careful clinical observation.

Presbycusis (or age-related hearing loss-ARHL) is defined as a progressive, bilateral, symmetrical sensorineural hearing loss, often impacting earlier higher frequencies - carrying critical information for speech understanding- sometimes with a partial conservation of low frequencies. ARHL is the third most frequent chronic diseases in the elderly population, immediately after cardiovascular diseases and arthritis, affecting more than 40% of the population over 65 years of age (Covelli et al., 2015). Furthermore, the prevalence of clinically significant hearing loss increases rapidly with age, passing from 3% in the population aged between 20 and 29 years, reaching almost 50% of subjects between 60 and 69 years old and affecting more than 80% of the so-called "old-elderly" (>85 years). It occurs as a result of a progressive deterioration of organ of Corti's hair cells with a progressive inability to stimulate the auditory nerve. Progressive deterioration of hair cells is long-life and it may occur as a consequence of both environmental and genetic factors, including cumulative effects of noise-exposure, ototoxic medication, DNA damages, reduction of mitochondrial function, vascular alteration and a reduced elasticity of cell membranes. Concerning genetic factors in ARHL, a number of genes seem to play a role (Huang & Tang, 2010); moreover, it might be an age-related increase of reactive oxygen species (ROS). ROS attack and destroy DNAm blocks of hair cells and connective tissue, causing a progressive reduced bio-energic activity of hair cells. Damages of connective tissues are also widespread in ARHL: reduction of outer and inner cochlear hair cells, progressive stiffening of basilar membrane, depletion and progressive degeneration of auditory cells to central auditory pathways up to cortical regions.

Along these factors, significant damages might be also inflicted by cumulative effects of external and psychosocial risk factors (such as lifetime exposure to noise, prolonged use of caffeine, alcohol and cigarettes, ototoxic medication, metabolic factors) and age-related predisposing clinical conditions (e.g. presence of diabetes, cardiovascular problems, degenerative and/or metabolic diseases, renal impairment, etc.).

Anyway, from a holistic point of view, effects of aging and contextual HL, go beyond the quantitative reduction of cochlear hair cells and subsequent reduction of perceived signal intensity. Rather, aging and HL are a complex set of deterioration process involving both peripheral and central processing. As a consequence, in typically ARHL, patients experience a consistent difficulty in speech conversation intelligibility in noisy environments and often present aberrant patterns of growth in loudness as sound intensity increases. This phenomenon, known as recruitment, is often related to a dynamic range that is narrowed by a low ceiling of tolerance for high-intensity sounds. This aspect may add further difficulties to understand conversation in challenging conditions.

As a matter of fact, nearly 60% of people over 65 years suffer from the so-called Auditory Processing Disorders (ADP) (Covelli et al., 2015). ICD-10 has recently classified Auditory Processing Disorders as specific processing dysfunctions along the central auditory nervous system, including both acquired and congenital form (Iliadou et al., 2017). This disorder, also defined as “Central Auditory Processing Disorder” is codified in the current tenth version of the International Classification of Diseases as H93.25 and it will be present in the forthcoming beta eleventh version. APDs may have detrimental effects on psychological wellness, determining anxiety, low esteem, and depressive mood. These disorders may interfere with learning, communication, social and emotional aspects of life

(Iliadou et al., 2017). Persons affected by ADP report symptoms such as difficulties in speech comprehension in challenging situation, “cocktail party syndrome”; easy distractibility, difficulties in repeating or recalling similar sounding words, problems in localization of sounds, difficulties in frequencies discrimination and in separation of auditory foreground from auditory background.

Whether or not ADP are present, presbycusis requires to consider additional clinical issues with aging central auditory pathways. Getting older, a reduced ability to discriminate verbal communication in terms of processing speed is one of the issues that occurs most frequently. Beyond this, ARHL entails a decline in quantity of cues critical for recognition of words; a poorer discrimination of frequencies, intensity and duration of sounds; a dysfunctional binaural central processing, which is critical in recognizing spoken language in a noisy environment, for localization and orientation to sound (Moore, 2016). As mentioned above, troubles following conversation in a noisy environment represents a typical manifestation of ARHL; moreover, preservation of low-pitched frequencies may enhance perception of background noise with worsening effect on speech perception. In old age, a reduced perception of Interaural Phase Difference (IPD) -the difference in the phase of a wave that reaches each ear- causes alteration of waveform; moreover, in case of a sensorineural HL, deterioration of fine temporal structure processing and altered discrimination of frequencies overlap across an auditory complex situation (Moore, 2016).

In addition, presbycusic patient typically presents a deteriorated processing of fine temporal structure, leading to reduction of temporal and spectral contrast, essential for preserving voice informative cues. The presbycusic patient therefore classically presents a discrepancy between the ability to hear sounds and the ability to understand them, especially in noisy environments.

Over the last 20 years, technological progress in audio-prosthetic solutions and provision of diagnostic-therapeutic programs have highly impacted and intervention in hearing impaired subjects (Gaylor et al, 2013; Pronk et al, 2011; Isaacson, 2010) leading to timely diagnosis and customized prosthetic solutions, more and more adapted to personal and audiological needs.

The Hearing Aid (HA) is a medical prosthesis designed to correct mild / moderate / severe hearing loss. Modern hearing aids are made up of analog or digital electronic devices, and the elements that constitute them are essentially three: a microphone, an amplifier and a receiver. The microphone converts the sound into an electrical signal which is amplified and then routed to the receiver, an acoustic speaker, which transforms it back into sound. The resulting sound, amplified up to a thousand times (60 db), is carried into the ear canal. Like all medical prostheses, hearing aids must also be designed in response to the functional needs and characteristics of the individual patient's pathology, or in any case adapted to them. However, in the elderly individual, some characteristics such as the compression of the dynamic field are strongly linked to residual cognitive abilities and the presence of other associated pathologies. To give an example, the wide dynamic fields, although they increase the audibility threshold, lead to a reduction of the spectral and temporal contrasts of the sound, resulting in more "fuzzy" sounds (Gatehouse et al., 2003).

In case of severe / profound hearing loss, when benefit from hearing aids is poor or absent, cochlear implant (CI) provides the most suitable and valid solution, offering an auditory threshold in the free field of 20-35 dB for the frequencies from 350 to 8000 Hz and an excellent understanding of speech in acoustically quiet environments.

A key element for a successful cochlear implantation is represented by the flexibility of stimulation parameters; flexible parameters allow personalized programs giving the opportunity of the best possible- use of CI.

In case of a monaural listening condition, when sound comes contralaterally, head constitutes a significant acoustic barrier with consequent difficulty following a conversation, understanding the verbal message and in localization and orientation to sound (Schafer, 2011; Zhang, 2013). As a matter of fact, the summing effect induced by two auditory input (from left and right ear) may provide an improvement loudness sensation in presence of noise up to 3 dB. Thus, restoration of bilaterality is to be preferred, ensuring better listening in noise, better lateralization and possibility of orientation: it is worthful to point out that binaural hearing, besides being critical for speech comprehension in noise, is crucial for localization and orientation to sounds. As mentioned above, troubles following conversation in a noisy environment represents a typical manifestation of ARHL; moreover, preservation of low-pitched frequencies may enhance perception of background noise with worsening effect on speech perception. As a matter of fact, a challenge posed by ARHL treatment relates to preservation of low-frequency residual hearing thresholds in combination with the provision of electric stimulation for the high-frequency hearing range (Skarzynsk et al., 2014).

The evaluation of the cost / benefit issues, the preservation of low frequencies that guarantee a good perception of the fine time structure and of F0 are the main reasons why the bimodal approach (CI / HA) in recent years is preferred over bilateral (CI / CI). It is in fact necessary to consider how the current CI technology offers a good perception of the waveform (envelope), to which the recognition of phonemes is linked, while it is unable to adequately transmit the fine temporal structure, well preserved in the HA and which is linked to the perception of F0, the appreciation of timbre and music (Wilson, 1997). The bimodal approach has in fact shown to offer better results in language perception especially in noisy environments (Ching et al., 2006; Cullington and Zeng 2011; Dincer D'Alessandro et al., 2018a; Dorman and Gifford, 2017). In recent years, numerous researches seem to confirm the significant benefits of a bimodal stimulation approach, regardless of the age of the subject and whether it is a quiet or noise condition. e.g., Ching et al., 2006; Devocht et al., 2017; Dincer D'Alessandro et al., 2015; Dorman and Gifford, 2017; Firszt et al. 2019; Hoppe et al., 2018; Hua et al., 2017).

4.1.2 Bimodal cochlear implantation in elderly patients

Abstract

Objective: Bimodal stimulation is a standard option for asymmetric hearing loss in adults. Questions have been raised whether receiving two stimulations may conflict in elderly listeners where the central integration of an acoustic/electrical signal may be very important to obtain benefit in terms of speech perception.

Design: Clinical retrospective study.

Study sample: The outcomes from 17 bimodal cochlear implant (CI) users were analysed. The test material consisted of speech audiometry in quiet and in noise (STARR and Matrix).

Results: Bimodal PTA and speech perception both in quiet and in noise were significantly better than CI or HA alone. Age showed a significant effect on bimodal STARR outcomes. Similarly, bimodal STARR scores improved significantly in comparison to Better Ear.

Conclusion: Both Matrix and STARR tests were very difficult for many elderly CI listeners from the present study group, especially in unilateral listening condition. The performance improved significantly, emphasising a good integration of acoustic and electric hearing in this group of elderly bimodal listeners. Overall results highlighted how a specific study, based on speech perception in noise in the elderly listeners, might shed light on the effect of speech test modality on bimodal outcomes.

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Key words: Older adults; bimodal hearing; cochlear implant; speech perception; adaptive test

Introduction

Bimodal listening involves a combination of two different stimulation modes: electrical stimulation via a cochlear implant (CI) in one ear and acoustic stimulation via a conventional hearing aid (HA) in the contralateral ear. As a result of the broadening of CI indications to include patients who demonstrate considerable residual hearing in the contralateral ear and who benefit from conventional amplification in that ear, bimodal listening is now becoming increasingly adopted (Ching et al., 2006; Sheffield & Gifford 2014; Devocht et al., 2017). Since existing CI technology conveys degraded spectrotemporal acoustic cues, in particular for the low frequency domain, the additional low frequency information provided by HA use in the contralateral ear has been shown to lead to improved speech perception, especially in the presence of noise (Ching et al., 2006; Cullington and Zeng 2011; Dorman & Gifford, 2017; Dincer D’Alessandro et al., 2018a). Indeed, an increasing number of patients, including elderly patients, are adopting bimodal hearing.

Several studies have now shown the benefit of bimodal stimulation in both adults and children, and in quiet and noise (Ching et al., 2006; Dincer D’Alessandro et al., 2015; Devocht et al., 2017; Dorman & Gifford, 2017; Hua et al., 2017; Firszt et al. 2018; Hoppe, Hocke & Digeser, 2018). Hence, the current recommendation is that all CI users should wear an HA in the contralateral ear unless there is clear evidence to suggest that this will have a negative effect on auditory perception (Cullington & Zeng, 2011; Dorman & Gifford, 2017; Firszt et al., 2018).

Most of these studies confirm the benefit of bimodal stimulation in terms of speech perception scores compared to CI alone but none have considered the role of aging, which is known to negatively affect auditory performance (Mosnier et al., 2014). In fact, in the elderly population, the central decoding process is affected by aging, and central integration of an acoustic/electric message may be important to gain a benefit in terms of speech perception (Moore et al., 2014).

The studies carried out in elderly cochlear implantees make use of a wide range of speech assessment tools such as the Hearing in Noise (HINT) (Zwolan et al., 2014), the Consonant- Nucleus-Consonant (Friedland et al., 2010; Roberts et al., 2013) and the AzBio sentence tests (Mahmoud & Ruckenstein, 2014), but all these studies performed these tests either in quiet or in noise with a fixed signal-to-noise ratio (SNR) but not with their adaptive mode (available in HINT). However, adaptive tests have long been recognised as a significant development in assessing benefits with auditory devices for two main reasons. First and foremost, such tests represent a more realistic acoustic scenario, as in everyday life, the level of speech and noise may vary rapidly from moment to moment; second, they allow outcome comparisons over time or across conditions by avoiding floor and ceiling effects (Plomp & Mimpen,1979).

The two newly developed tests in Italian, the Sentence Test with Adaptive Randomised Roving level (STARR) (Dincer D'Alessandro et al., 2016) and the Matrix test (Puglisi et al., 2015), are based on an adaptive SNR paradigm. Both tests measure the speech recognition threshold (SRT) where 50% of sentences are repeated correctly, and therefore both are considered useful to determine speech understanding in more realistic conditions compared to speech tests presented with a fixed SNR paradigm. They have been recently used in the evaluation of cochlear implantees and results reflected the great difficulty that CI users face in challenging listening situations. Even the better CI performers including bimodal users could not achieve performances similar to those from people with normal hearing (Boyle et al., 2013; Dincer D'Alessandro et al., 2018a; Dincer D'Alessandro & Mancini 2019; Gallo & Castiglione 2019). However, so far a specific evaluation has not been carried out in elderly CI users.

The STARR and Matrix tests differ in their adaptive paradigms and in their degree of semantic predictability. The Matrix test is based on a closed-set speech perception task and uses sentences with semantic unpredictability, with either primary signal or noise that is adaptively varied (Kollmeier et al., 2015). On the other hand, the STARR test uses everyday sentences that are semantically predictable, while both speech and noise signals vary in level (Dincer D'Alessandro et al., 2016).

In the literature, it has been argued how semantic content might be critical when assessing speech recognition in elderly participants. Different speech and cognitive tests may highlight distinct aspects of listening and engage cognitive processes to different degrees (Heinrich, Henshaw & Ferguson, 2015; Hua et al., 2017), inevitably influencing the degree of compliance of elderly listeners to testing (Gordon-Salant & Cole 2016). More specifically, the semantic complexity of the speech material for the Matrix test may be influenced by working memory capacity (Hua et al., 2017) and consequently may underestimate the amount of bimodal benefit (Gordon-Salant & Cole 2016). The STARR performance may depend to a lesser extent on working memory and because of the semantically more predictable speech material, may allow elderly CI listeners to make use of cognitive compensation and successfully complete the speech perception task. On the other hand, the STARR test is known to be challenging for CI listeners due to testing at varying speech levels (low-, medium- and high-level speech). In particular, testing at low-level has been shown to deteriorate significantly the STARR performance in CI users (Boyle et al., 2013). Indeed, such an effect might be even stronger for elderly CI listeners.

The principal aim of the present study is to examine whether elderly listeners show bimodal benefit for speech perception and whether patients' age influences the amount of bimodal benefit. A further aim is to gain insights into the use of the STARR and Matrix tests with different adaptive paradigms as well as into the role of semantic predictability of the speech material in elderly listeners.

Material and Method

Participants

Seventeen patients with bimodal hearing were enrolled in the study. Mean age at testing was 73 ± 1.2 years (range: 65–84) whilst mean age at implantation was 67 ± 1.7 years (range: 52–81). Duration of deafness was, on average, 30 years with only two patients having a duration shorter than 5 years (range: 2–60 years). All patients were consistent HA users in the contralateral ear, both before and after cochlear implantation. In the CI ear, 15 participants wore an HA before implantation: 6 used it up to surgery and 9 had not used it for a period ranging from 0 to 3 years before surgery. Almost all of the patients had a long history of hearing loss with a mean period of HA use before CI of 20 ± 4.8 years.

All patients had severe to profound hearing loss on the CI side: pre-op unaided low frequency (125–500 Hz) pure tone audiometry (PTA) ranged between 25 and 120 dB HL (mean 76.6 dB HL). Four participants showed mean aided sound field PTA in the range

125–500 Hz ($SF\text{-}PTA_{125–500\,Hz}$) of 48 ± 4.1 dB HL. In the contralateral HA side, mean aided $SF\text{-}PTA_{125–4000Hz}$ was 55.3 ± 4.4 dB HL (46 ± 4 dB HL at frequencies 125–500 Hz and 64.6 ± 6 dB HL at frequencies 1000–4000 Hz). The mean word recognition score (WRS) in quiet for bilateral HA listening condition before implantation was 63 ± 8%. Post-operative average $SF\text{-}PTAs_{125–4000Hz}$ were 36.0 ± 1.37 dB

HL and 29.5 ± 1.6 dB HL for CI alone and bimodal listening conditions respectively. Average $SF\text{-}PTAs_{125–500Hz}$ were 38.5 ± 1.6 dB HL for CI alone and 30.7 ± 1.8 dB HL for bimodal listening, while average $SF\text{-}PTAs_{1000–4000Hz}$ were 34.8 ± 1.8 for CI alone and 28.4 ± 1.7 dB HL for bimodal listening.

Mean period of CI use was 4.9 ± 1.1 years (range: 1–15). Seven patients used an AB HiRes90K implant together with a Naida speech processor fitted with a HiRes Optima sound processing strategy; 10 patients used a Med-El Concerto or Syncrony implant together with a Sonnet speech processor fitted with a FS4 or FS4-p strategy. Both CI and HA microphones were set in omnidirectional mode, and accessory filters were disabled. HAs were Siemens Signia SP7, Widex Unic 440 Fusion or Phonak Naida that were fitted with NAL-NL1, Widex fitting rationale or Phonak adaptive prescriptive formulas, respectively.

Procedures

The present study is a clinical retrospective chart review of all adult patients who were implanted at Cochlear Implant Centre, University Sapienza – Policlinico Umberto I – Rome. Inclusion criteria for study enrolment were age ≥65 years and consistent use of an HA in the contralateral ear. Data collection included age at implantation, gender, side of implantation, CI model, duration of hearing loss (years of severe-to-profound sensorineural hearing loss, established by tonal and speech audiometry and considering their last year to use a telephone), aetiology, HA use in the implanted and in the contralateral ear, speech perception assessment in both quiet and noise as described below. As for the neurological history of patients, the Mini-Mental State Examination (MMSE) (Magni et al. 1996) was used to exclude patients with any cognitive or psychological problems or dementia.

Ethical approval was obtained by the local Institutional Review Board. This observational study was carried out in accordance with the ethical requirements of the Helsinki Declarations, the Epidemiological Good Practice Guidelines of the ICH (International Conference of Harmonization), and the existing legislation in Italy.

Both HA and CI fittings for individual recipients were controlled before testing. Most comfortable levels were balanced and confirmed to be appropriate when listening bimodally to avoid any discomfort due to a loudness summation effect. Prior to achieving balancing, each CI/HA side was first checked for loudness perception with a loudness scaling procedure at 250, 1000 and, where appropriate, 4000 Hz by using the A§E psychoacoustic test suite (Vaerenberg et al., 2011). This procedure was followed by a loudness balancing as recommended by Ching, Hill, and Dillon (2004).

As all patients were tested in unilateral and bimodal listening conditions, when testing the poorer ear (PE), the device in the better ear (BE) was removed and the ear was plugged with soft silicon earplugs (Senner GmbH, Leipzig, Germany) and muffed with Sennheiser HDA 200 supra-aural audiometric earphones (Sennheiser Electronic Corporation, Old Lyme, CT, USA). Unaided and aided PTAs at octave frequencies between 125 and 8000 Hz were obtained using frequency-modulated tones in a standard soundproofed booth. Assessment was performed through an Aurical audiometer (Otometrics Taastrup, Denmark) connected to TDH39 headphones or to a loudspeaker placed at 0 azimuth at 1 m distance from the participant's head when testing in SF. Speech and noise stimuli were presented via a computer and a preamplifier connected directly to a single loud- speaker which was placed at 0 azimuth and at 1 m distance from the participant's head. Speech recognition was assessed in quiet and in noise with an adaptive SNR paradigm, in each listening mode with randomised presentation: CI alone, HA alone and bimodal.

Speech perception

Speech perception in quiet was assessed with the disyllabic balanced words lists from Italian Speech Audiometry (Turrini et al., 1993), and with the speech signal presented at 0 from the participant's head at 65 dB SPL.

Speech perception in noise was evaluated using the STARR (Dincer D'Alessandro et al., 2016) and Matrix tests (Puglisi et al., 2015) in Italian.

The STARR test was developed to obtain a reliable SRT assessment with varying signal levels so as to test the effects of CI parameters such as Automatic Gain Control compression or noise-reduction algorithms on speech recognition in deaf patients wearing an auditory prosthesis (Boyle et al., 2013). The Italian adaptation made use of sentences from the standard Italian speech recognition test (Cutugno, Prosser & Turrini, 2000; Dincer D'Alessandro et al., 2016). The corpus consisted of 10 test lists, each containing 15 sentences, all recorded with a male voice. For competition, speech-shaped noise was used. Three presentation levels (50, 65 and 80 dB SPL) were used for sentences, with five presentations at each level within a single test list. The number of words in each sentence ranged from 3 to 7, and for each sentence, three key words were used for scoring. Lists were presented in noise resembling the long-term spectrum of speech. The spectrum level was flat up to 800 Hz and then dropped at approximately 12 dB per octave. After presentation of a sentence produced by a given speaker, the participant was asked to repeat it as accurately as possible. For the response to a sentence to be scored as "correct," at least two of the three key words had to be repeated back correctly, otherwise the response was scored as "incorrect." After a correct score, a more unfavourable SNR was used for the next sentence while, after an incorrect score, a more favourable SNR was used for the next sentence. The initial SNR was 20 dB and varied adaptively following the participant's response, by adjusting the noise level, keeping the speech level at 50, 65, or 80 dB SPL. The same SNR was used across all three speech presentation levels. The SNR step size started at 10 dB; dropped to 5 dB after the first reversal of the adaptive track and dropped again to 2.5 dB after a further reversal. The SRT for each test list was computed by averaging the SNRs for the last nine sentences together with the SNR at which a next sentence would have been presented. The STARR SRTs ranged from 10 to 125 dB SNR. Previous studies in CI listeners have shown that in the STARR test, 20 dB SNR was considered to be the cut-off threshold between better and poorer performers (Dincer D'Alessandro et al., 2018a, 2018b). This was because the STARR test asked a listener to achieve a score greater than 67% correct for a given SNR so that the next SNR would become more adverse and the adaptive track would converge. Listeners with a score 67% correct at the initial SNR (20 dB), were also able to respond sensibly to SNR manipulation. Their performance tended to improve with a more favourable SNR and to deteriorate with a more adverse one, allowing this adaptive test to estimate the 50% point on their psychometric curve. Thus, their SRTs revealed a significant ability for speech perception in noise and SRT increases within this threshold (20 dB) reflected a performance deterioration for speech perception in noise. On the other hand, for poorer listeners, SRTs reflected their difficulties in understanding speech even when there was no detectable competing noise due to very high SNRs, especially for sentences presented at 50 and 65 dB SPL. Moreover, listeners with very high SRTs (poorer than 60 dB) could not achieve a score 67% correct even at the maximum sentence presentation level of 80 dB SPL.

Despite such previous STARR findings showing limitations to the use of the STARR test in poor performers (Boyle et al., 2013; Dincer D'Alessandro et al., 2018a, 2018b), the present study did not exclude poor performers who could not successfully complete the test in unilateral listening mode. This would not allow the study to track performance changes in poor CI/HA listeners and to differentiate progress for those who were just beginning to be able to manage the STARR test because of bimodal benefit. Moreover, findings would not reflect overall elderly bimodal listeners.

Matrix was developed to be compatible with other matrix tests (Kollmeier et al., 2015) to allow cross-language comparisons. The test lists can be used for accurate repeated measurements with the same listener as they use semantically unpredictable sentences with a fixed syntactic structure (name-verb-number-noun-adjective; e.g. *Sofia trascina poche matite utili*, which is Italian for “Sophie drags a few useful pencils”) and a random selection of items. The Matrix test is a closed-set test in nature due to a limited number of item options. However, in contrast to everyday sentences, those generated in the Matrix test have low semantic predictability. Because of the semantically unpredictable structure, the lists cannot be memorised easily and thus, can be used repeatedly. Various approaches exist for the execution of such tests. The SRT can be measured with either noise or speech level fixed at 65 dB SPL and in a broad range of individuals with normal or impaired hearing in an unaided or aided condition. In this study, assessment was performed with an open-set response format using speech noise fixed at 65 dB SPL. Each test list was composed by 30 sentences and was preceded by two training lists. Each correctly repeated word was recorded and scored (word scoring). According to the number of correctly understood words in the preceding sentence, the software adapted the speech level for the next sentence, and starting at 0 dB SNR, the procedure iterated an estimate of the SNR level where 50% of presented words were understood.

The test-retest reliability was 0.7 dB for the Italian STARR test (Dincer D’Alessandro et al. 2016) and 0.5 dB for the Italian Matrix test (Puglisi et al. 2015). Both SRT procedures com- menced after explaining the task to the participants and after presenting a training list for the STARR test (Dincer D’Alessandro et al. 2016) and two training lists for the Matrix test (Kollmeier et al. 2015) following the guidelines from previous studies for learning effects. The participants were asked to repeat the sentence as accurately as possible and were told that not every word needed to be correct. The participants were rec- ommended to ask for a break whenever needed to avoid per- formance deterioration due to fatigue. Two STARR lists giving a total of 30 sentences were administered when testing each listening condition, with the aim of increasing the reliability in the outcome comparisons.

Statistical analysis

All statistical analyses were performed with IBM SPSS Statistics for Windows (v. 22.0, IBM Corp., Armonk, NY, USA). Non-parametric statistical tests were carried out for analysis since a Shapiro–Wilk test indicated that the data were not normally distributed for STARR performance from HA alone listening ($p < 0.001$). The percent correct responses for speech perception in quiet were transformed to Rationalised Arcsine Units (RAUs) to avoid the ceiling effect (Studebaker, 1985).

Considering physical limitations such as noise in the booth and electrical noise in the amplification system, an SNR ceiling value of 30 dB was set for participants' scores that were above 30 dB at the STARR test as well as for those with no response at the Matrix test. A Friedman test was performed for comparison between the three listening conditions (CI alone, HA alone, bimodal). Post-hoc analysis was performed using Wilcoxon tests with the Bonferroni correction. The effect size was calculated using Rosenthal formula r Z/ N (small effect = 0.10-0.30, moderate effect = 0.30-0.50 and large effect ≥ 0.50) (Rosenthal, 1994). Differences between BE and bimodal listening were compared using a Wilcoxon test. The CI/HA side with the better results at STARR and Matrix tests was considered as the BE. Spearman rank-order correlations were used for bivariate correlations between variables. The cut-off level for statistical significance was set to 0.05.

Results

Detailed descriptions of demographics and audiological outcomes are reported in Tables 1 and 2 (all Supplementary data cited but non included in this paragraph related to this article can be found at https://doi.org/10.1080/14992027.2020.1843080).

The BE was the CI side in 9 patients and the HA side in 7 patients. It was not possible to identify the BE for one participant who did not achieve a measurable performance in bimodal listening condition neither for the STARR nor for the Matrix tests.

The effects of listening mode

SF-PTAs125–500Hz and SF-PTAs1000–4000Hz differed significantly between the three listening conditions (p < 0.001). Post-hoc analysis showed that CI and HA SF-PTAs125–500Hz were not significantly different (p =0.124, r =-0.26). Differences were statistically significant for SF-PTAs1000–4000Hz (p < 0.001, r=-0.62). SF-PTAs from the bimodal listening condition were significantly better in comparison to those from both CI (SF-PTAs125–500Hz: p = 0.002 and SF-PTAs1000–4000Hz: p = 0.001, r= -0.54); and HA alone (SF-PTAs125–500Hz: p < 0.001, r= -0.60; and SF-PTAs1000–4000Hz: p < 0.001, r= -0.62). SF-PTAs were not significantly different between BE and PE (SF-PTAs125–500Hz: p =0.266, r= -0.19; SF-PTAs1000–4000Hz: p = 0.124, r=-0.26), while for the bimodal listening condition, because of the summation effect, PTAs were always significantly better than either BE or PE (r values ranged from -0.55 to -0.62 and p values ranged from 0.001 to < 0.001). Bimodal outcomes were significantly better than those of both CI alone (p = 0.003, r=- 0.52), and HA alone listening conditions (p= 0.002, r=-0.52) with a large effect size. CI alone outcomes were not significantly different from those of HA alone (p =0.929, r= -0.01). Taking 20 dB SNR as the cut-off threshold between poorer and better performers (Dincer D'Alessandro et al., 2018a; 2018b), the analysis of outcomes has shown how the overall group had reasonable outcomes with bimodal hearing. Twelve out of 17 participants had a score better than 20 dB SNR with bimodal hearing, while only 6 versus 7 participants were able to show such a score for the CI and HA alone listening conditions, respectively. Only one participant had a bimodal score that was significantly poorer than the CI score [a performance difference greater than 2 dB SNR as described by Dincer D'Alessandro et al. (2016)]. Bimodal listening provided a significantly improved performance in comparison to the BE (p =0.027, r=-0.38) with a moderate effect size. Eleven patients had STARR scores better than 20 dB SNR with their BE.

The Matrix performance differed significantly between the three listening conditions (p =0.001). Outcomes for the bimodal listening condition were significantly better than those of both CI alone (p= 0.017, r=- 0.40) and HA alone listening conditions (p < 0.001, r=- 0.60) with a small and large effect size respectively. CI alone outcomes were not significantly different from those of HA alone (p =0.551, r= - 0.08). Bimodal listening did not render a significantly improved performance in comparison to BE (p=0.121, r =- 0.27). Fifteen out of 17 participants had a score better than 20 dB SNR with bimodal hearing, while only 9 participants were able to show such a score for the CI and HA alone listening conditions.

In patients with complete sets of measurements, STARR and Matrix findings were significantly correlated (r_s =0.79, p < 0.001). Elderly participants who were better STARR performers (an SNR better than 20 dB) showed a median Matrix SRT 9.1 dB (range 6.3 to 11.5 dB) for CI alone, 5.4 dB (range 3 to18 dB) for HA alone and 5.4 dB (range -0.5 to 23 dB) for bimodal listening condition.

The effects of demographic factors

Statistical analysis showed that the age of the participants was significantly correlated with STARR scores for bimodal listening condition (r_s=0.49, p =0.046). Hence, older patients performed worse on the STARR test when listening bimodally. Hearing loss in either the right or the left ear was not correlated with age (r_s=0.23, p = 0.390 and r_s=0.14, p = 0.590, respectively). Duration of CI use was positively correlated with CI alone WRS in quiet (rs=0.54, p =0.025) whilst the duration of hearing loss did not reveal any significant correlations with outcomes (p > 0.05). For the Matrix test, the correlations with demographic variables were not statistically significant (p > 0.05).

Discussion

Aging has been shown to play an important role in the speech perception skills of CI users, and the literature reflects the significant performance deterioration in elderly listeners (Mosnier et al., 2014; Beyea et al., 2016; Yang & Cosetti, 2016). For this assessment, previous studies made use of tests that were performed either in quiet or in noise presented with a fixed SNR (Friedland et al., 2010; Roberts et al., 2013; Mahmoud & Ruckenstein, 2014; Zwolan et al., 2014; Beyea et al., 2016; Yang & Cosetti, 2016). To the best of our knowledge, no studies have been published so far aimed specifically at evaluating the outcomes of bimodal listening in the elderly based on an adaptive methodology. Hence, the present study focussed on the bimodal benefit in elderly cochlear implantees, measured with two recently developed speech tests with an adaptive SNR paradigm, the Italian STARR (Dincer D'Alessandro et al. 2016) and the Italian Matrix (Puglisi et al. 2015).

The overall results from the present study showed that auditory perception in elderly participants improved significantly with bimodal listening when compared to unilateral mode, even in respect to the performance of the BE. Such findings indicated that elderly listeners were able to integrate the electric and acoustic cues provided by the CI and the HA to at least some degree. For elderly participants with long lasting hearing loss, one important issue might be that, after years of having to process asymmetric inputs, the neural circuitry might have been affected by plasticity so that time and training might be needed before experiencing bimodal advantages. Successful CI rehabilitation, among various factors, might depend on the extent to which the brain can reclaim or optimise auditory processing networks after implantation (McKay, 2018). Therefore, all patients participating in the present research had a minimum bimodal hearing of 1 year, with an average of 4.9 years. Such duration of experience provided the time necessary for most of them to adapt to the new listening mode and to benefit from it.

The age of the patients showed significant effects on bimodal listening performance for the STARR test, even in the group composed purely of elderly listeners. Hence, older patients performed worse on the STARR test. The significant negative effects of age were further supported by comparing the present findings with those reported in the literature for a younger population. Here in elderly listeners, there was a tendency towards poorer speech recognition in quiet, especially when listening with CI alone (an average of 60% in the present sample vs 78% in younger CI listeners) (Dincer D'Alessandro et al., 2018a). The differences were even more remarkable for the STARR test (an average of 30 vs. 14 dB for CI alone and 11 vs. 8 dB for bimodal listening) (Dincer D'Alessandro et al. 2018a) and the Matrix test (an average of 13 vs 4 dB for CI alone and 8 vs 3 dB for bimodal listening) (Gallo & Castiglione 2019). The present data were in agreement with recent literature in which the SRTs in noise ranged from -2 to +20 dB SNR (average SRT=+8.1, ±7.1) in CI users aged between 58.3 and 93.9 years (Claes et al., 2018). In comparison, in younger adult CI listeners some implantees were able to perform very well (between 0 and 5 dB SNR for LIST in noise) while others performed relatively poorly (van Wieringen & Wouters, 2008). Duration of deafness did not seem to significantly affect the results of bimodal stimulation in the present sample. However, it should be noted that, in the present study, the average duration of hearing loss was 30 years and only two participants had a duration less than 5 years. This fact and the small sample size might have contributed to such outcome differences. A recent study in a larger population of Italian speaking CI listeners with a larger variability in the duration of deafness has shown that this had a significant effect on STARR performance (Dincer D'Alessandro et al., 2018a). Similarly, a large retrospective collection of data from 2251 patients implanted since 2003 in 15 international centres showed how durations of both moderate and profound hearing loss before implantation were significantly correlated with postoperative auditory performance in quiet and in noise (Lazard et al., 2012). On the other hand, the duration of CI use was positively correlated with CI alone WRS in quiet, even in a group of participants with at least 1 year of CI experience. Such results were in line with recent studies showing a perceptual improvement for CI users with poorer performance even after 1 year (De Seta et al., 2016; Dincer D'Alessandro et al., 2018b). Concerning SF thresholds, participants in the present study showed a CI-SF average of 36 dB HL and the bimodal condition significantly contributed to improve overall audibility over the monolateral condition, implying a bilateral summation effect, reaching an average audibility level of 30 dB HL in the range 125–8000 Hz, slightly worse than what has been reported in the literature in younger patients (Potts et al., 2009). Higher SF levels in elderly participants when compared to younger CI wearers have already been reported by Aimoni et al. (2016) and Benatti et al. (2013). These studies showed an average PTA125–8000 Hz ranging from 36 to 44 dB HL in elderly CI wearers. One possible explanation for higher CI thresholds in the elderly population could be a decreased tolerance to electric loudness, partly dependent upon poorer auditory processing for frequency and loudness discrimination when compared to younger participants (He, Dubno, and Mills 1998; Moore 2014). In fact, both monaural and binaural auditory processing deteriorate with increasing age, even when audiometric thresholds are within the normal range (Moore 2014). Despite these premises, CI listeners in the present study group have been shown to benefit from the summation effect, where both CI and HA contribute to improve overall bilateral audibility. This fact might have contributed to the significant performance differences for the STARR test, which was previously found to be indicative of perception of the low-level speech presented at 50 dB SPL. In fact, Boyle et al. (2013) reported significantly poorer performance for the low level speech than for the medium-level speech in CI users, and Dincer D'Alessandro et al. (2018a) observed a significant effect of CI PTAs on the STARR performance even for a group of CI listeners with PTAs better than 40 dB HL. Indeed, an inability to perceive speech at the 50 dB SPL level appeared to be the major factor limiting the STARR results, especially in poor CI performers (Boyle et al., 2013; Dincer D'Alessandro et al., 2018b). In this sample of elderly listeners, the speech perception score in quiet for bimodal hearing was significantly better than for the CI alone or HA alone conditions. Although bimodal scores in quiet were similar to results from several studies carried out in younger adult populations, the CI alone outcomes tended to be poorer as mentioned above (Dincer D'Alessandro et al., 2018a). It has been reported that one of the factors influencing the benefit of combined electric and acoustic hearing was the amount of residual hearing in the low frequency range below 500 Hz. It was described that for bimodal fitting to yield significant benefits, hearing loss in the contralateral ear should not exceed 80 dB HL in the low frequency range ≤ 500 Hz (Illg, Bojanowicz, & Lesinski-Schiedat, 2014). Such outcomes might even have had indications for the inevitability of bilateral implantation whenever bimodal listening seemed not to be beneficial (Luntz, Yehudai & Shpak 2007; Illg, Bojanowicz & Lesinski-Schiedat 2014). Also, an acoustic hearing threshold < 90 dB HL for octave frequencies 1000 and 2000 Hz in the non-implanted ear was shown to be important for sentence recognition in noise (Neuman & Svirsky, 2013) while a threshold < 100 dB HL was associated with perception of interaural time difference (ITD) and to lateralisation capability (Francart, Brokx & Wouters, 2009). Participants from the present study showed, on average, residual serviceable hearing for octave frequencies lower than 2000 Hz. This fact has significantly contributed to improve speech perception through bimodal listening. Although there is a greater role for hearing acuity on word recognition in quiet (Humes, 2005; Van Rooij & Plomp, 1990), in the elderly population, a better speech perception in quiet has also been positively correlated with the degree of cognitive function (Heydebrand et al., 2007). Hence, it cannot be excluded that the bimodal listeners in the present study group might have benefitted from improved availability of auditory cues to support the cognition and attention processes. Similarly, with previous studies in bimodal recipients (Potts et al., 2009; Devocht et al., 2017; Dincer D'Alessandro et al., 2018a; Gallo & Castiglione, 2019), speech perception in noise was significantly improved because of the availability of bilateral cues. In fact, with the addition of low frequency acoustic hearing in the ear contralateral to the CI in bimodal fittings, there was a significant improvement, especially for sentences in noise (Dorman & Gifford 2017). In this study, all participants but one were able to achieve 50% recognition in bimodal listening mode for both STARR and Matrix tests, indicating an ability to understand speech in the presence of noise. However, even the best bimodal listeners did not reach a level of performance similar to that reported in people with normal hearing. On the other hand, when tested with CI alone or HA alone, several participants could not achieve an SRT for the Matrix test. Likewise, their STARR scores were very poor. Although differences in STARR performance between bimodal and HA alone were not significant, it was noteworthy how an improvement in the bimodal listening condition was observed in most participants, even when the CI alone or HA alone side was providing poor speech recognition especially in noise. In these patients, the perception of low-level speech was probably the main factor leading to performance differences between the best performers and those with poorer performance. For listeners with a score poorer than 30 dB SNR, SRTs reflected rather their difficulties in understanding speech even when there was no detectable competing noise due to very high SNRs, especially for sentences presented at 50 and 65 dB SPL. Boyle et al. (2013) observed how the poor performance at 50 dB SPL might have worrying implications for real-life communication, for which speech levels can fall well below 65 dB SPL (Pearsons, Bennett & Fidell, 1976). The role of the BE in the bimodal listening condition has recently been discussed by Hoppe et al. (2018). That study showed a reduction in the percentage of participants with bimodal gain in the order of 10–20% points once the reference was made to the BE. The authors speculated that a study addressing a bimodal condition should refer to the BE, regardless of modality, in order to evaluate the true bimodal effects of the second ear with the complementary modality. In the present study group of bimodal elderly participants, the BE showed a similar HA/CI distribution in the study group. BE-PTA was still significantly different from bimodal-PTA, emphasising that, in this specific group of elderly participants, the PE was still contributing to auditory perception, resulting in a significant performance improvement for the STARR. Bimodal speech perception at STARR test was significantly different in comparison to the BE, emphasising a good integration of acoustic and electric hearing in this group of elderly bimodal listeners. The perception at low-level speech specifically measured at STARR test might have contributed to reveal statistically significant differences that were not observed at Matrix test. Hence, the improvement was conceivably provided by high frequency perception from the CI and low frequency perception from the HA. Past research has shown how participants with moderate–severe high frequency hearing loss might have benefitted from high frequency speech cues when these were made audible up to 4000 Hz (Weatherby, Henshall & McKay 2003; Simpson, McDermott & Dowell, 2005). If residual contralateral hearing was adequate, the addition of a contralateral HA, besides significantly contributing to hearing outcomes, partially compensated for the negative hearing fluctuations as well as for the slow initial progress with the CI (Luntz, Yehudai, and Shpak 2007; Illg, Bojanowicz, and Lesinski-Schiedat 2014). On the other hand, the hearing preserved at low frequencies, which was typically addressed by HA amplification, was associated with improved low frequency pitch and speech perception in noise. In a previous work by Dincer D'Alessandro et al. (2018a), low frequency pitch discrimination was poorer than what was reported in the literature for normal hearing people, and it was shown to be significantly improved in bimodal listeners. In the same subgroup of bimodal users, the improvements in PTA125–1000 Hz, pitch discrimination and STARR performance also improved significantly in bimodal listening mode when compared to the CI alone condition.

A further aim of this study was to assess the feasibility of speech material with different complexities in elderly participants where the central integration of an acoustic/electrical signal might be very important (Moore 2014). Together with the presence of competing noise which exerts an influence on working memory and attention engagement (Neher et al. 2009), the complexity of the target speech signal might also influence the relationship between cognition and speech recognition (Neher et al. 2009; Baskent, 2010;

Saija et al. 2014; Hua et al. 2017). Research focussed on elderly participants has shown how intelligibility tests can vary depending on their auditory and cognitive demands and their sensitivity to the challenges that auditory environments pose on functioning. For instance, Heinrich, Henshaw, and Ferguson (2015) reported that using linguistically more complex speech material such as sentences led to a stronger relationship between cognitive ability and speech-in-noise performance compared to a phoneme-discrimination task. The authors suggested that practitioners and researchers should think carefully about the objective outcome measures they choose as different speech and cognitive tests will highlight different aspects of listening and engage different cognitive processes (Heinrich, Henshaw & Ferguson, 2015). Indeed, the results of the present study have shown how elderly participants could make use of different types of acoustic and electric inputs when challenged with different SRT speech material, as the participants' performance improved in both STARR and Matrix tests when assessed in the bimodal condition. The STARR and Matrix tests were highly correlated although they were intrinsically different from each other. The tests differed in their adaptive paradigms and in their degree of semantic predictability. The Matrix test was based on a closed-set speech perception task but used sentences with semantic unpredictability. Instead, the STARR test used everyday sentences that were semantically predictable but varying speech and noise levels might have been more challenging than speech level alone that varied adaptively at Matrix test (Boyle et al. 2013; Kollmeier et al. 2015; Dincer D'Alessandro et al. 2016). On the other hand, the Matrix test was characterised by a higher number of items/lists, hence a longer time of execution (Puglisi et al. 2015). By analysing two groups of elderly participants aged <70 and ≥70 years, Rohloff et al. (2017) reported how only 49.6% and 53.2%, respectively were able to complete the Matrix test. Furthermore, all participants required more time to become acquainted with the test set-up and execution. The authors also suggested that "diminished intellectual and cognitive abilities may bias hearing results when the hearing tests were complex and required more attention". Indeed, the participants in the present study group might have benefitted both from the semantically meaningful stimuli in the STARR test and the shorter time of execution. Overall results highlighted how a specific study, based on speech perception in noise in the elderly listeners, might shed light on the effect of speech test modality on bimodal outcomes.

Limitations of the study

One possible limitation of the study was the variability in participants' HA characteristics which might have influenced the results. Nevertheless, independently from prescriptive formulas, gain was adjusted to obtain a well-balanced electric/acoustic SF-PTA, which was shown to be significantly improved compared with the single side alone condition. A further limitation was the lack of a deep understanding of the cognitive status of our patients before the cochlear implantation. The Mini-Mental State Examination (MMSE) was administered to exclude severe cognitive deficits and dementia before testing, but no testing was adopted to measure the working memory which would be linked to speech perception performance in noise. A further prospective study should be carried out to assess the effects of working memory on the Matrix and STARR performance in elderly CI users.

Conclusion

Elderly participants affected by asymmetrical severe/profound hearing loss were shown to benefit from bimodal stimulation in speech recognition tasks in both quiet and noisy conditions. In most cases, access to partial bilateral cues may generate measurable benefits in terms of speech perception. Both Matrix and STARR tests were very difficult for many elderly CI listeners from the present study group, especially in unilateral listening condition. Indeed, several participants showed SRTs above 30 dB SNR when listening with HA or CI alone. However, the tests were more amenable to elderly listeners when listening bimodally. The performance improved significantly, emphasising a good integration of acoustic and electric hearing in this group of elderly bimodal listeners. On the other hand, age showed a significant negative effect on bimodal listening STARR performance, even in the group composed purely of elderly listeners. Similarly, bimodal speech perception was significantly different in comparison to the BE only at STARR test. Aspects such as word familiarity, sentence length, syntactic structure as well as testing at various speech levels might have contributed to reveal statistically significant differences that were not observed at Matrix test. In particular, the poor performance at low-level speech might have implications for real-life communication where speech levels can fall well below 65 dB SPL. Traditional speech perception testing may underestimate this effect, and outcomes from varying speech levels and noise may help to improve CI fitting.

4.2 Aging in hearing loss: cognitive and psychological correlates

A moderate to severe hearing loss may have a significant impact on quality of life of an old person. It is necessary to consider consequences first of all in terms of reduced speech perception and comprehension, with daily and constant hampered and depleted communication. The lower and lower motivation to communicate may finally impede participation in social life, relational, cultural and aggregation activities. That, in turn, might trigger worsening effects on psychological and /or neuropsychiatric clinical conditions, such as cognitive deterioration, incident dementia and depressive conditions. In old age, biological, cognitive, psychological, and social factors may assume new meanings and different relevance compared to younger ages.

All these aspects should be carefully addressed in treatment and care of hearing-impaired elderly patients. Moreover, presence of overlapping problems related to aging of central auditory pathways, vascular and degenerative pathologies, cognitive deterioration, depressive or anxiety mood, are all aspects adding clinical complexity that should not be overlooked.

4.2.1 Presbycusis and depressive mood

In the last decade, several studies highlighted possible links between presbycusis and depression, although studies show mixed results. Gopinath et al. (2009) conducted a three years long prospective cross-sectional study on a sample of 1328 subjects aged over 60 years, finding a higher incidence of depressive symptoms in bilateral hearing impaired (HI) subjects than normal hearing (NH) peers. Interestingly, HI women younger than 70 years had five time higher odds to receive diagnosis of depression than NH peer women. This association was not found for women older than 70. Moreover, regardless of age and gender, HI subjects that reported using a hearing aid for a minimum of 1 hour per day had a significantly lower odds of depressive symptoms than non and non-frequent users. The finding of a higher probability of depressive symptoms in HI women than men is consistent with previous studies (Kvam, Loeb, & Tambs, 2007; de Graaf & Bijl, 2002) reporting more mental problems in deaf women than in deaf men. De Graaf & Bijl (2002) investigated mental health conditions separately in a large sample of prelingual and postlingual adults with severe-profound HL, reporting the higher rates of mental distress in postlingually deaf women (43%). These rates were higher than prelingually deaf women (32,4%), prelingually and postlingually deaf men (27.1% and 27.7%, respectively) and than general NH population (men: 22.0%; women: 26.6%). For both prelingual and postlingual HI categories, the risk of mental distress was higher in those with more communication problems, lower levels of self-esteem, and poorer acceptance of hearing loss.

In a cross-sectional study, Contrera et al. (2016) investigated association between ARHL and emotional vitality in a sample of 1903 HI elderly aged 75-86 years, finding a significant correlation between presbycusis and low scores in emotional vitality, defined as having a high sense of personal mastery, happiness, low depressive symptomatology, and low anxiety. Contrera et al. (2016) found that participants with moderate or severe hearing loss had 23% lower odds of emotional vitality than NH and peer with mild HL. Unlike Gopinath et al. findings (2009), in this study no significant association emerged between the use of hearing aids and better outcomes of emotional vitality.

Different results were obtained by Boi et al. (2012), and by Mener, Betz, Genther and Lin (2013): in both studies an association between use of hearing aids and lower rates in depressive symptom was found. Mener, Betz, Genther and Lin (2013) in a nationally representative study of older patients, analyzed association between presbycusis, use of hearing aids and diagnosis of major depressive disorder in a sample of 1029 adults aged 70-79 years: independent association between use of hearing aids and lower odds of diagnosis of major depression and any depressive symptom was found. However, Mener et al. (2013) underlined the impossibility of determining direction of this association. For example, they wondered if the very same presence of depression may lead a person not to use hearing aids or, from another point of view, subjects with normal mood are more likely to require rehabilitation treatments; otherwise, it may be possible a cause-effect direction, such as: use of hearing aids, improving listening skills, may increase elderly social and relational participation hence leading to reduction of depressive symptoms. Similar conclusions have been reported by Boi et al. (2012): Authors conducted a study on 15 HI subjects aged over 70 years suffering from presbycusis and comorbid depressive symptomatology. A reduction in depressive symptoms and a significant improvement in quality of life were observed early on with the use of hearing aids.

4.2.2 Presbycusis and cognitive decline

In recent years, a growing number of studies seem to suggest the existence of an association between presbycusis and deterioration of cognitive functions (Gallacher et al., 2012; Lin et al., 2011; Lin, Metter et al., 2011; Lin, Yaffe et al., 2013; Quaranta et al., 2014). As pointed out by numerous researches (eg., Cherko, Hickson, & Buttha, 2016; Wayne & Johnsrude, 2015; Mudar & Husain, 2016) it is still not possible to define a unique cause-effect link neither to define direction of this connection.

A body of research suggests deafness as cause of a faster cognitive decline, leading to extra effort at the sensory-perceptual level with negative consequences to cognitive, attentional and mnemonic resources (Tun, McCoy &Wingfield, 2009).

According to the original definition of cognitive resources (Kahneman, 1973) it refers to a limited set of attentional reserves that must be allocated among several mental operations. Consequently, the more resource demanding a mental operation is, the fewer resources will be available for use elsewhere in the system (e.g., Craik & Byrd, 1982).

Some authors suggested that cognitive decline in hearing impaired adults may be a consequence of an overinvestment of brain activity on auditory processing and language comprehension functions, resulting in a significant detriment for other cognitive processes. The effect of hearing loss on cognitive load is suggested by studies demonstrating that in difficult auditory conditions, cognitive resources are sent on perceptual processing with detriment of other cognitive processes (Pichora-Fuller, Schneider, & Daneman, 1995; Wingfield & Grossman, 2006). With aging, sensory functions together with several perceptual and cognitive functions tend to decline and slow down. Nevertheless, in normal aging, language comprehension typically remains well preserved. Wingfield and Grossman (2006) reviewed data from functional magnetic resonance imaging (fMRI) to describe a two-component model of sentence comprehension: a core sentence-processing area located in the perisylvian region of the left cerebral hemisphere, and an associated network of brain regions underpinning working memory and other executive functions needed for comprehension of long or syntactically complex sentences. Authors used this two-components model to describe the nature of brain compensatory recruitment, suggesting that, also in elderly person, this plasticity in neural recruitment contributes to the stability of language comprehension in the aging brain (Wingfield & Grossman, 2006). So, in case of age-related hearing loss, the more cognitive processing resources must be spent for processing auditory informations, the fewer resources available for non-perceptual cognitive processes remain (Tun, McCoy, &Wingfield, 2009). As a matter of fact, hearing impaired adults show poorer performance than NH peer on high demanding working memory tasks, even after controlling for age and abilities to understand verbal instructions (Rutherford, et al., 2018; Schneider, Li & Daneman, 2007). This detrimental effect might be instantaneous at one end (*information-*

degradation hypothesis) or resulting in slow gradual brain plasticity changes at the back (*sensory deprivation hypothesis*) (Mudar & Husain, 2016).

According to the "Common Cause Hypothesis" (Linderberg and Baltes, 1994; Anstey, Luszcz & Sanchez, 2001) presbycusis and cognitive impairment could be the expression of the same neurodegenerative process. So, sensory functioning could be a strong late-life predictor of individual differences in intellectual functioning and seen as indicator of the physiological integrity of the aging brain. Although it is not possible to totally reject this hypothesis, it is necessary to underline how, from a neurological point of view, the perception of pure tones (PTA) – as universally used audiological test - is a measure of peripheral auditory processing. Pure tone audiometry is considered to be a measure of the auditory periphery because detection of pure tones does not involve high-level cortical processes, relying on cochlear transduction and neuronal afferents to brainstem nuclei (Pickles, 2008): in fact, even subjects with severe cognitive impairment seem to show normal results in these audiometric measures (Lin, Metter et al., 2011). Neurological studies on Alzheimer Disease (AD) have not found neuropathological correlates in the peripheral auditory pathways (Lin, 2011). On the contrary, researches focused on central auditory processing disorders and risk of dementia seem to confirm a strong association between the two conditions, with significant correlations between low scores on the dichotic listening tests and dementia diagnosis, in particular, for Alzheimer Disease (AD) (Gates et al., 1996; Gates, Beiser, Rees, D'Agostino, & Wolf, 2002; Gates, Anderson, McCurry. et al., 2011). In conceptual framework of the "Common Cause Hypothesis", some authors have argued the existence of a common neurobiological process such as inflammation, vascular disease, or hereditary factors. An example is offered by people with the gene variant APOE4 (apoliprotein E4). Recent researches have shown that people with APOE4 have higher levels of amyloid proteins, with a higher risk for AD (van der Lee et al., 2018). This gene variant may cause an alteration in metabolism of lipids in neurons and astrocytes, significantly depleting microglia activity to remove dying or damaged neurons, pathogenic and waste substance (O'Grady, Boyles, Sper, Deruyter, Strittmatter, & Worley, 2007).

From a quite different angle, hearing loss may unmask or accelerate cognitive decline in elderly, by depleting cognitive reserve. Cognitive reserve is a latent construct theorized to account for the discrepancy between observed brain deterioration and ultimate clinical outcomes (Stern & Barulli, 2019); it represents the brain's ability to temper consequences of pathologic damages by using pre-existing cognitive resources or by enlisting compensatory processes. Decreased cognitive reserve, as indexed by educational level, occupational attainment, or participation in leisure activities, is associated with significantly increased risk of dementia. As a matter of fact, epidemiological studies suggest that lifelong educational, social, occupational and leisure experiences can increase this reserve (Stern, 2012). The Author suggests to divide the concept of reserve into two types: "*brain reserve"*, which refers to differences in the brain structure that may increase tolerance to pathology, and "*cognitive reserve"*, which refers to differences between individuals in how tasks are performed: this might enable some people to be more resilient to brain changes than others. "Greater understanding of the concept of cognitive reserve could lead to interventions to slow cognitive ageing or reduce the risk of dementia" (Stern, 2012, p. 1006)

Another strand of studies suggest a possible predisposition to hearing loss in subjects suffering from the so-called Mild Cognitive Impairment (MCI), defined as a slight impairment in cognitive function without a severe dysfunction in daily activities (Levey, Lah, Goldstein, Steenland & Bliwise, 2006) and without a necessary progression in dementia or AD (Petersen, 2004).

Despite the actual inability to characterize a precise cause-effect relationship between HL and cognitive decline, several studies have pointed out the association between the two conditions. For example, studies that used functional and structural magnetic resonance imaging and electroencephalography (Peelle et al, 2011; Wong et al., 2010; Eckert et al, 2012; Lin et al., 2014), observed anatomical and functional alterations - independent of age, sex and educational level - in adults with presbycusis. Peelle et al. (2011) conducted a study using voxel-based morphometry, a neuroimaging analysis technique that assess focal differences in brain anatomy, using the statistical approach known as parametric statistical mapping. Authors observed the existence of an association between presence of hearing loss and a reduction in the volume of grey matter in the primary auditory cortex. Peelle et al. (2011) also found a significant correlation between high frequencies (>2000 Hz) hearing loss and a reduction in bilateral cortical grey matter volume. According to researchers, these results would suggest that even a slight-moderate hearing loss may lead to chronic dysregulations of neural activity in speech processing, with a depletion of grey matter in the primary auditory cortex. Similar findings have been found by Eckert et al. (2012) and from Wong et al. (2010). In particular, Wong et al. (2010) found a correlation between a reduced ability to speech perception in noise and a reduction in grey matter volume and density in the prefrontal cortex. Interestingly, this reduction was not found in younger hearing-impaired subjects, even after controlling the effect of age on brain volume. Analogous results have also been obtained by Husain et al. (2011) and by Lin et al. (2014), thus supporting the hypothesis of the existence of a link between presbycusis and a more rapid decline in the volume of cerebral grey matter. Several studies have documented cognitive decline in individuals with hearing loss in both auditory (e.g., verbal memory) and non-auditory cognitive tasks (e.g., Digit Symbol Substitution Test) (see Mudar & Husain, 2016 for a review), suggesting that effects of age-associated hearing loss on cognition are central in origin. As underlined by Mudar & Husain (2016), understanding whether these cognitive declines are driven by additional neuroplastic changes and the impact of ARHL on neural structure and functionality might have both theoretical and clinical implications, for example, enabling researchers to assess the efficacy of hearing aids remediation approaches and determine whether their effects are permanent (Lin et al., 2013 in Mudar & Husain, 2016).

Among available researches on the relationships of ARHL and dementia. one of the most robust body of studies is that conducted by Lin, Metter, et al. (2011) within a large research project known as the Baltimore Longitudinal Study of Aging (BLSA): data from 639 subjects aged between 36 and 90 were analyzed in this study, all without signs of dementia or cognitive decline in a baseline condition, followed for 17 years. All participants were administered a battery of audiometric, neurological, and neuropsychological tests. Findings highlighted a significant positive correlation between degree of hearing loss and risk of dementia: through multiple regression techniques analysis, Lin et al. (2011) analyzed the hazard ratio corrected for confounding factors (age, sex, educational level, vascular diseases): the hazard ratio for incident dementia was double in subjects with mild hearing loss, triple in case of moderate deafness, even reaching a hazard ratio of 4.9 in subjects with severe hearing loss. Moreover, correcting the model for age, sex and other confounding factors, for hearing loss >25dB the risk of dementia would tend to increase linearly with severity of hearing impairment.

In a further study Lin (2011) investigated association of hearing loss with cognition in a sample of 605 subjects aged 60-69 years who underwent both standardized audiometric and cognitive assessment. Through an exploratory analysis, Author found a cross-sectional association between increasing HL and decreasing cognitive functions, measured by the Digit Span Substitution Test (DSST), a nonverbal measure of executive function and psychomotor speed.

Even after adjusting for age, sex, and hearing aid use, greater hearing loss was significantly associated with lower DSST scores (Lin, 2011). These findings have been confirmed by a further study (Lin et al., 2013) in which 1984 older adults, without cognitive impairment at baseline, were followed up for 11 years in cognitive and hearing functions. In total, 1162 individuals with baseline hearing impairment had annual rates of cognitive and executive functions decline that were 41% and 32% greater, respectively, than those among individuals with normal hearing with a risk for incident cognitive impairment linearly associated with the severity of baseline hearing loss.

Anyway, despite promising scientific advances in the field of the study of cognitive functioning in ARHL, it is not currently possible to univocally define the precise nature of their association. In Table 4.1, some possible non causal mechanisms that might explain the observed association between hearing loss and cognitive decline are shown.

1.	Hearing impairment might influence neuropsychological testing more than cognition per se
2.	Poor verbal communication associated with hearing loss may confound cognitive testing
3.	Upstream common causes with no conditions causally related to other
4.	Greater sensitivity of tests in one domain (hearing or cognition) could identify deficits in that domain prior to the other, leading to the appearance of an illusory causal relationship.
5.	Hearing impairment may introduce a systematic bias into neuropsychological assessments, many of which were designed and validated using verbal explanations of instructions and/or presentation of stimuli
6.	Cognitive screening tools may capture only limited variability in a normally aging population, with ceiling effects that could potentially lead to an underestimation of the true relationship between age-related hearing loss and cognitive decline.
7.	Hearing loss brings older adults to medical attention more frequently (overdiagnosis)

Table 4.1 Possible non causal mechanisms underlying the link between age-related hearing loss and cognitive decline

4.3 Towards a biopsychosocial approach to age-related hearing loss. Exploring quality of life in deaf adults: what are we looking for?

All above permits us to catch the potential significant impact of ARHL on quality of life. Summing up, it is necessary to consider its consequences in terms of reduced perception and communication that inevitably has consequences on participation in social life, cultural aggregation activities and relations. That, in turn, might trigger worsening effects on psychological and /or neuropsychiatric clinical conditions, such as the risk of premature cognitive deterioration, incident dementia and depressive conditions.

Over the past twenty years, research in treatment of severe-profound deafness in adult and elderly patients have increasingly highlighted the need for multimodal and integrated interventions, able to target both audiological and extra-audiological variables related to presbycusis. In other words, it is pointed out the need to extend focus of the intervention beyond the mere correction of the hearing deficit (Kricos, Holmes, & Doyle, 1992; Hickson & Worrall, 2003; Boothroyd, 2007).

The necessity to assume an integrated perspective on the concepts of health, disease and disability has been strongly pointed out by the International Classification of Functioning, Disability and Health (ICF) (World Health Organization, 2001).

The ICF, as a conceptual frame of reference adopted by the World Health Organization, underlines how the concept of "health" should be conceptualized from a multiple perspective, first of all by identifying the interactive effects on personal global functioning. In this sense, the ICF gets to consider "disability" and "functioning" as interactive and dynamic constructs made up by individual health conditions and contextual/ environmental factors.

The ICF proposal has represented an important shift from the "old medical approach" to the new "biopsychosocial approach". In the previous classification system (International Classification of Impairments, Disabilities, and Handicaps) (ICIDH) the terminology used to define health-related conditions highlighted the individual deficit: impairment, disability, handicap. In that conceptual framework, however, a first significant semiological distinction was proposed between the concepts of impairment (what does not work properly), disability (what cannot be done due to impairment) and handicap (the negative impact of disability on quality of life).

In the current ICF classification system (World Health Organization, 2001) terms such as operation, activity and participation replaced the previous missing-oriented terminology. The shift is from the individual who "does not work" to the interaction between individual characteristics and environmental characteristics with an inclusion of positive and functional elements.

Disability results from the interaction of multiple variables: therefore, it can vary significantly from individual to individual. Functioning and disability of a person should therefore be conceived as a dynamic interaction between health conditions (diseases, ailments, injuries, traumas, etc.) and contextual factors.

Below a summary of ICF system organization is provided.

ICF is organized in two main parts:

Part one (Functioning and Disability): The components of Functioning and Disability can be expressed in two ways. On the one hand, they can be used to indicate problems (e.g. impairments, limitation of activity or restriction of participation, grouped under the term disability umbrella); on the other they can indicate non-problematic (neutral) aspects of health and the states related to it, grouped under the umbrella term "operations". These components of functioning and disability are interpreted through four separate but related constructs.

Body functions and structures can be classified through changes in physiological systems or anatomical structures. The Activity (execution of a task or an action by an individual) and Participation (involvement in a life situation) components are classifiable through two constructs: Ability (ability to perform tasks in a standard environment)) and Performance (possibility to perform tasks in the current environment).

Part two (the Contextual Factors): it includes both personal and environmental factors (attitudes, the physical and social environment in which people live and lead their existence). Environmental factors interact with all components of functioning and disability facilitate or hinder personal functioning. As a consequence, functioning and disability are therefore conceived as a dynamic interaction between health conditions (diseases, ailments, injuries, traumas, etc.) and contextual factors.

More specifically, problems impairing personal functioning are categorized through three interconnected domains:

- impairments: problems in body functions and structures; for example, conditions of deafness, blindness, paralysis;
- - activity limitations: difficulty in carrying out activities, for example, hearing, seeing, walking;
- - restrictions on participation: problems experienced in social involvement, for example, going to the cinema or attending a party.

In this conceptual framework, the term disability is intended to refer to the difficulties that an individual may encounter in each or all areas of functioning and originates from the continuous and unavoidable interaction between the objective health condition and environmental, contextual and personal factors.

Thereby, this biopsychosocial approach is going to be the conceptual framework in treatment and care of hearing-impaired elderly. By applying the ICF table to the condition of severe-profound hearing loss in the elderly, a schematic representation of the relationship between the different variables could therefore be represented as follows (Figure 4.2):

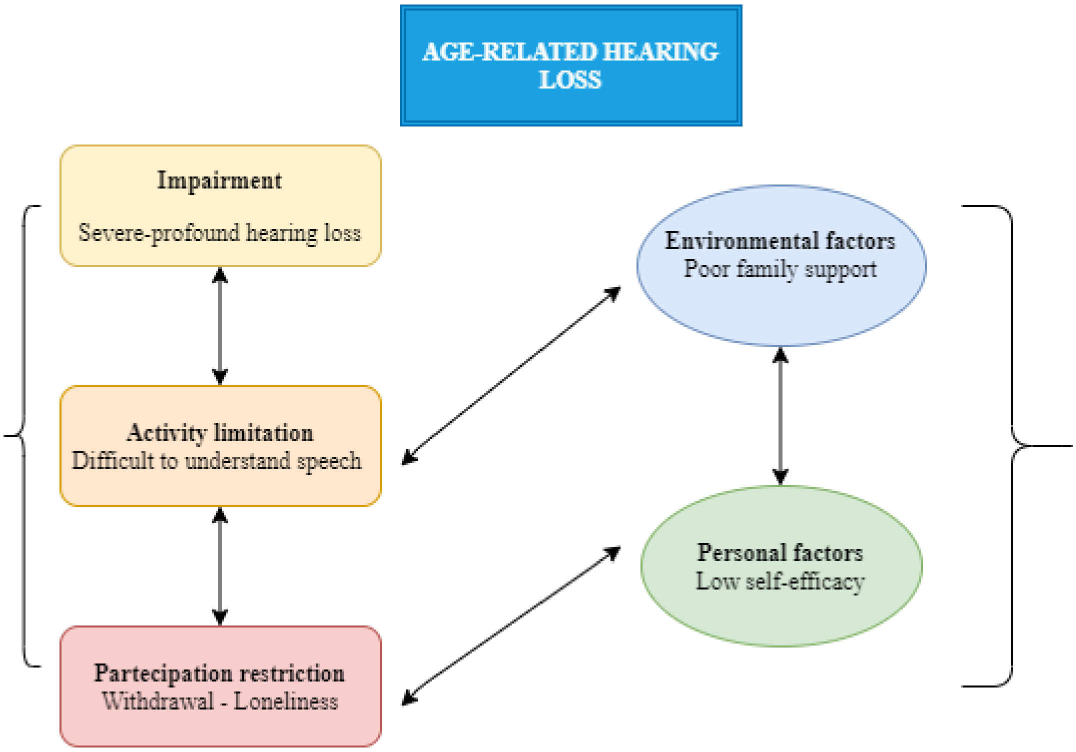

Figure 4.2: Application of the ICF approach to ARHL

Application of the ICF approach is leading clinicians to consider the importance of multimodal approach to the hearing-impaired elderly patient, focusing on audiological, communicative, cognitive and psychological aspects. Final goal should be a concrete improvement in quality of life through a reduction of limitations imposed by sensory deprivation.

We should consider disability within an interactive, holistic and ecological framework including physical impairments, activity limitation, participation restrictions, social and personal factors (WHO, 2001): thus, the auditory restoration may not be all it takes for addressing psychological, emotional and relational consequences of deafness (Boothroyd, 2007). This holistic, multi-component point of view on disability should invite clinicians to broaden their focus on the interventions, including measures of psychosocial, cognitive and relational consequences of hearing impairment. In other words, rehabilitation should represent a holistic intervention to person and not just the management of a sensory impairment.

Moreover, many elder hearing aid users, despite a daily and constant use of hearing amplification, continue to describe a great impairment in communication and daily social participation that negatively affects their quality of life (Hickson & Worrall, 2003).

Kushalnagar et al. (2014) conducted open-ended semi-structured interviews with adults born or become deaf early in life: topic was how being deaf influenced their lives across several situations and life context. Authors' aim was to delineate a conceptual model of health-related quality of life (QoL) in deaf adults, with particular attention to congenital and early-onset HI subjects. Using a coding method of thematic analysis, an Adult-QoL Deaf Model was conceptualized (Fig.4.3).

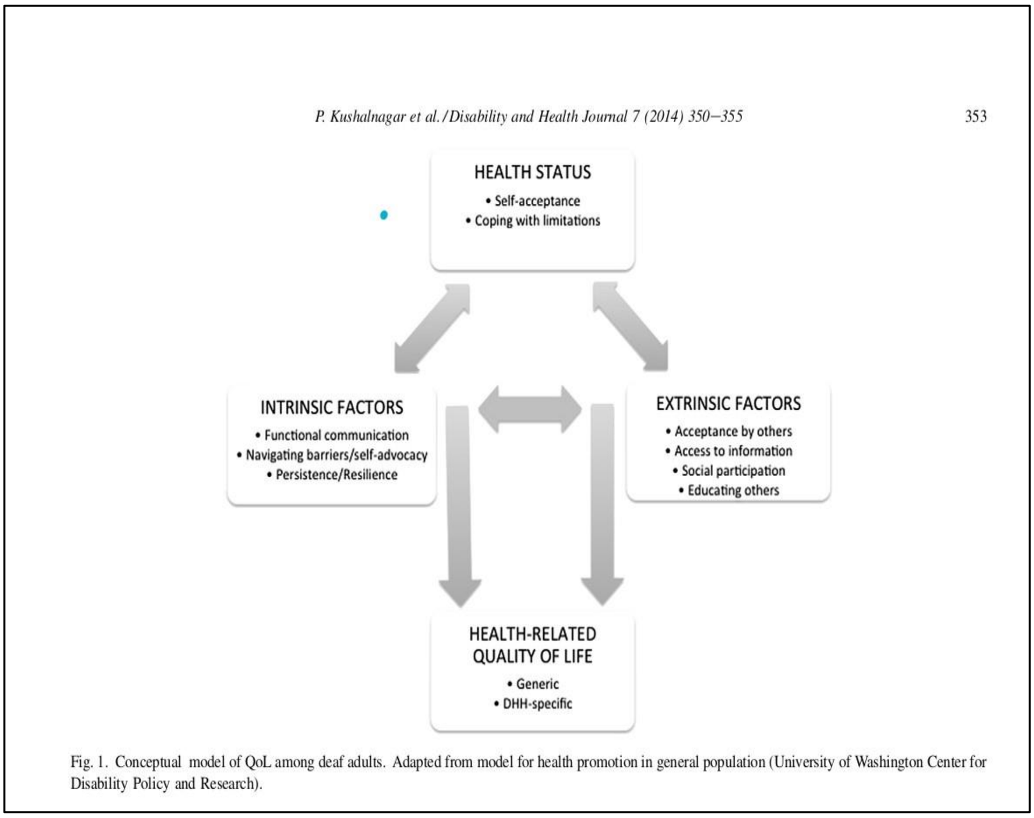

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Fig. 1. Conceptual model of QoL among deaf adults. Adapted from model for health promotion in general population (University of Washington Center for Disability Policy and Research).

Fig. 4.3 Conceptual model of QoL in adult hearing loss (Kushalnagar et al. 2014)

The A-QoL Deaf Model delineates the relationship between:

Health status: across all ages, good physical health and good mental health status were recognized as important components of deaf-specific QoL. Similarly, to general population, self-perception of good health status is a factor highly affecting positive QoL outcomes. Interestingly data analysis showed that self-acceptance of hearing loss plays a large part in health status self-perception and it is the basis for recognition of communication limits and communication skills. In turn, to recognize and accept limitations imposed by deafness provides the basis for coping with hearing and communicative challenges. Coping with limitations associated with being deaf is another factor that contributes to health status.

Intrinsic Factors: to rely on functional communication skills emerged as an important contributor to perceived QoL. In the presented conceptual model it represents an important component for the perceived ability to adapt to communication and social challenges, solve problems, and overcome barriers.

Extrinsic Factors: the necessity of being recognized and accepted in their environment resulted a consistent theme across all ages. Social participation, feeling included in group conversations and others' disposability to adapt communications to their needs were identified as important aspects of QoL in deaf adults.

Analysis of the data in this study suggests that self-acceptance of hearing loss is an important part of the person's health status and QoL. The adoption of the modern International Classification of Functioning, Disability and Health (ICF) (World Health Organization, 2001) approach over recent years has been leading clinicians to consider the importance of a multidisciplinary approach to the hearing impaired elderly patient, focusing on audiological, communicative, cognitive and psychological aspects. In this way, as we'll see in the following final chapter, the ultimate goal of multimodal interventions is essentially that of concretely improving quality of life of hearing-impaired persons, reducing communicative, social and emotional limitations imposed by sensory deprivation.

Chapter 5. Towards a Scaffolding Approach to Rehabilitation for Cochlear Implant elderly persons

5.1 Introduction: effectiveness of additional or alternative interventions to hearing aid fitting on quality of life and psychological status in elderly

Hearing loss is the most common sensory impairment in elderly and one of the most challenging disabilities in the aged population, with a great number of consequences for quality of life, psychological wellness, functional independence, and cognitive abilities (Kramer, 2005). Constant and repeated communication failures due to a hearing impairment may represent one of the most frustrating experiences in aged people. Moreover, the age-related health problems can negatively affect sense of independence and autonomy, causing daily restriction in social participation, feeling of sadness, isolation, inability, incompetence and depression. The acquired hearing loss might deplete psychological functioning and social interactions (Heine & Browning, 2002), increasing feeling of worthlessness, impotence, and isolation, with a consequent worsening of quality of life.

An increasing amount of research has been pointing out the necessity of adjunctive form of intervention, such as communication and psychological programs, in order to meet the various and complex needs of age-related hearing impaired subjects (Kricos, Holmes, & Doyle, 1992).

The multiple effects of an age-related hearing impairment need to be tackled beyond the only hearing aid fitting, through interventions, training, and programs on communicative and psychosocial needs of hearing-impaired elderly people (Rosenhall, 2001).

Anyway, treatments explicitly focused on elderly hearing-impaired people's psycho-social wellness and quality of life are quite rare.

A revision of literature has been carried out in order to appraise the impact of these kind of interventions on quality of life of HI elderly population, using PubMed and Web of Science until August 2019. We searched on databases combining the terms quality of life, psychological and psychosocial wellbeing with the terms "hearing-impaired" "deaf", "elderly" and "interventions" and their possible variations.

For inclusion, reports were required to consider the effect of an extra-audiological intervention on quality of life and psychological or psychosocial wellbeing of hearing-impaired elderly subjects. We considered only studies of samples with a mean age > 60 years.

250 citations were found, of which only 31 full text were screened as relevant for the study. Twenty-two studies were excluded because not respondent to the criteria. At the end of the selection process, we found nine studies whose outcome measures include these issues with interventions explicitly addressing to enhance quality od life and/or psychological wellbeing. Of these, just three (Kricos, & Holmes, 1992;Andersson, Green, & Melin,1997; Oberg, Bohn, Larsson, & Hickson, 2014) analyzed those outcome measure in a sample of only elderly, whilst the majority of these studies focused on hearing impaired adult population (also including those over 60 years).

For the goals of this minireview, the selected reports were divided into two main categories: *in-vivo* rehabilitation programs and self help/individual home programs.

- *In vivo rehabilitation programs*

Five studies focused on effectiveness of in vivo programs on quality of life and/or psychosocial/psychological wellbeing of elderly with hearing impairment were found.

Kricos and Holmes (1996) compared the effectiveness of two forms of rehabilitation for hearing impaired elderly: an Analytic Auditory Training and an Active Listening Training .The effectiveness of the programs was evaluated through a pre and post training assessment on a sample of 78 adults with a mean age of 70 years. All subjects were hearing aids (HA) users with a mild to moderate hearing loss. Twenty-six subjects were assigned to control group (no treatment); 26 to the Analytic Program and 26 to the Active Listening Training.

The Authors used the Hearing Handicap Inventory for the Elderly (HHIE) (Ventry &Weinstein, 1982) for assessing social and emotional effects of hearing impairment, and the Communication Profile for the Hearing Impaired (CPHI) (Demorest & Erdman, 1987) for assessing psychosocial functioning. Concerning results, neither the analytic auditory training group nor the active listening group showed a significant improvement over control group in the level of hearing handicap self-perception. On the other hand, for the Active Listening group only, a significant improvement over control group was found in several aspects of psychosocial status. In particular, Kricos and Holmes (1996) reported that the active listening group had an improvement in their ability to communicate effectively even in adverse listening environment through an enhanced use of verbal and nonverbal strategies. Moreover, the participants to this rehabilitation group showed an improvement in the subscale of CPHI called “personal adjustment”, including dimensions of acceptance of loss, attitude to others, withdrawal, and denial. According to Kricos and Holmes “…these improvements in personal adjustment could be attributed to the individuals learning coping strategies to help them deal with communication breakdowns” (1996, p.227). In this sense, they underlined the advantage of using an active listening training, considered more suitable than analytic training, for older adults.

An *in-vivo* program that focuses on quality of life and psychological status of older people with hearing impairment is the Active Communication Education (ACE), developed from a research program at the University of Queensland (Hickson & Worrall 2003).

Original version of ACE program was created to help adults with hearing loss to become more effective communicators, providing them with skills and strategies to cope with everyday difficulties. ACE program is intended both for HA users both for nonusers. The ACE handbook defines it as a program "to help adults with hearing loss to become more effective communicators and to provide them with strategies to cope with everyday difficulties" (Hickson, Worral & Scarinci, 2015, p. *V*)

Three studies exploring the effectiveness of ACE in elderly hearing impaired were found (Hickson, Worrall, & Scarinci, 2007b; Oberg, Bohn, Larsson, & Hickson, 2014; Oberg, Bohn, & Larsson, 2014).

Hickson et al. (2007b) conducted a double-blind randomized control trial involving 178 old adults with a mild/moderate hearing impairment. Of these, 46% did not have an hearing aid (HA) 13% had a unilateral HA and the remaining 41% bilateral HA. The experimental group undertook the ACE program only, whereas the control group undertook a five weeks placebo social program followed by the ACE. The Authors expected a greater improvement in communication, well-being, 5and quality of life for participants who completed the ACE program compared to the CG after the first 6 months. Furthermore, they expected a within group effect of completion of ACE after the social program in the CG.

The effectiveness of ACE on quality of life was measured by the SF-36 (Ware & Sherbourne, 1992): no improvements for the first hypothesis neither for the second one were found.

The psychological wellbeing was assessed by a short version of the Psychological Wellbeing Scale – PWB (Ryff, 1989). No improvements for the first hypothesis was found. For the second hypothesis, the authors found a consistent improvement for psychological wellness, communication participation and the reduction of activity limitation for participants who completed the ACE program. Nevertheless, the analysis of between-group comparison revealed an effect size of ACE not large enough to support the hypothesis of a significant difference. The Authors hypothesized that “better outcomes may be obtained with ACE if participants spend more time together in the group environment” (Hickson et al., 2007b, p. 226), since changes associated to ACE + SOCIAL were significantly better if compared to ACE only.

The second study on the effectiveness of ACE (Oberg, Bohn, Larsson & Hickson, 2014) was a within-subjects intervention with a three-time assessment: before ACE, three weeks post and six months follow-up. The main goal was to assess the effectiveness of a Swedish version of ACE in a in a sample of 23 older-old hearing-impaired adults (all aged 87 years) with moderate hearing loss, of whom 78% were HA users. Results were similar to those found by Hickson et al. (2007b), although outcomes measures were different. In Oberg et al. (2014) psychological wellbeing was assessed through the short form of the Geriatric Depression Scale (Yesavage et al., 1982), and quality of life was evaluated using the visual analogue scale (VAS) of Euroqol-5d (EuroQol Group, 1990). Descriptive statistics revealed no significant differences in quality of life and psychological wellbeing before and after the intervention at three weeks and six months follow-up. Moreover, very small changes were found in the within group effect size although there was an improvement in the use of communication strategies.

Oberg, Bohn & Larsson (2014) conducted a further intervention study on the effects of a modified version of ACE, with pre-, post-treatment and at six months follow-up assessments. The modified version of ACE included more psychological oriented contents and psychological exercises. The study group consisted in 67 hearing impaired adults - mean age 69 years – with mild to moderate hearing loss, of which 51 were HA users (74%). Concerning the outcome measures, quality of life was evaluated using the VAS of the Euroqol-5d (EuroQol Group, 1990) and the psychological status was assessed with the Hospital Anxiety and Depression Scale –HADS (Zigmond & Snaith,1983). The Authors found a significant long-term within-group improvement in the use of communication strategies and the levels of participation and psychological wellbeing. A significant increase in communication strategies was found in the older participants (>66years) both in the short and in long term as revealed by the post hoc analysis. These results agreed with entries from Oberg, Bohn, Larsson and Hickson’s (2014) previous study, showing a good effectiveness of ACE program on learning communicative skills in hearing impaired old subjects. Interestingly, nature of benefits changed with severity of hearing loss: 1) participants with mild hearing loss improved in activity and participation in pre, post and in six months follow-up; 2) participants with moderate hearing impairment improved scores in anxiety and depression in the long term. “People with more severe hearing loss might have a longer experience of hearing loss and may have already learned communication strategies compared with those with milder hearing loss” (Oberg et al., 2014, p.857).

Another type of intervention focusing on quality of life of hearing-impaired elders was implemented by Saunders and Forsline (2012). Authors compared the effectiveness of two forms of counseling on a sample of 74 adults with a moderate hearing loss on average and a mean age of 66 years. All participants worn HA for at least three months with scarce satisfaction. Researchers’ starting point was to overcome the too resource-intensive programs through implementation of a single-session counselling: main goals were to improve outcomes in HA in terms of better understanding and acceptance of hearing loss, use of communication strategies, and ability to explain hearing difficulties to others. Authors compared the effectiveness of a 30-minutes informational single session counseling (SSC) with the effectiveness of a 30- minutes performance perceptual counseling (PPT). Both types of counseling were delivered by a research audiologist and were based on a patient-centered communicative approach. The first type of counseling (SSC) focused on providing tips for hearing-aid and communication strategies, while the second (PPT) had a metacognitive approach, focusing on the discrepancy between objective and perceived ability to understand spoken language through hearing aids. Participants were randomly assigned to SSC or to PPT. The primary outcome measure was the Psychosocial Impact of Assistive Devices scale (PIADS) (Day & Jutai, 1996), a self-rating measure of quality of life in three domains: adaptability, sense of competence and self-esteem. Scores showed an improvement for participants, for all the three domains: no significant difference in outcomes between groups were found. So, the study seems to show effectiveness of a single-session patient-centered approach, but without significant difference between informational hearing aid counseling and meta-cognitive counseling.

- *Self-help manuals and individual home interventions*

Common ground of these self-help home programs is the possibility to offer interventions in a cost-effective way, fronting of the great economic and time burden of *in vivo* rehabilitative sessions. Andersson, Green and Melin (1997) study is an experimental between groups pre-post study involving 19 mild/moderate hearing-impaired subjects aged

67-75 years, all hearing aid users. The main purpose of their program was to motivate older hearing-impaired adults to take control of their hearing problem in a cognitive-behavioral psychological framework. Conceptual starting point was the psychological behavioral-cognitive oriented methods of counseling. Consistently they applied a behavioral treatment approach to hearing tactics: “the notion is to view hearing as being a behavior as well as a physical function” (Andersson et al., 1997, p. 523). Effects of Andersson’s behavioral treatment on psychological wellness were evaluated in terms of enhancement of coping skills, that are classically defined as the cognitive, emotional and behavioral skills a person is able to use for addressing life troubles (Folkman &Lazarus, 1985).

Self-Help Manual: Hearing tactics- the art of coping with hearing impairment (Green and Andersson, not published)	
Chapter one	How to applicate relaxation tactics
Chapter two	Hearing Tactics for an effective communication
Chapter three	Effective use of hearing aids
Chapter four	Problem Solving: cognitive strategies for facing challenges related to hearing loss

Fig. 5.1 Main contents of the “Hearing tactics” book (Green and Anderson, unpublished) (Andersson et al., 1997)

The Communication Strategies Scale (CSS) of the Communication Profile for Hearing Impaired (Demorest & Erdman, 1986, Demorest & Erdman, 1987) was used as main outcome measure: it is a five point response scale that assesses three types of communication strategies: “Verbal Strategies”, “Non-Verbal Strategies” and “Maladaptive Behaviour”. The verbal and non-verbal subscales include adaptive behaviours that compensate for the problems associated with hearing impairment (such as lip-reading, remaining silent and asking for repetition); the maladaptive behaviour subscale include behaviours that interferes with effective communication (such as pretending to understand, avoiding communication situations, trying to dominate conversations) (Helvik et al., 2007). Statistical analysis conducted for CSS scores revealed a significant treatment effect. Post-hoc analyses revealed significant difference between-groups on the verbal subscale total score and on of the CSS-

CPHI, revealing a better functioning in acting behavioral strategies to cope with hearing and communication difficulties.

An alternative way to offer additional communication programs in a cost-effective way consists in using internet for online rehabilitation trainings. Several studies on effectiveness of online education program for patients with chronic disease, tinnitus and anxiety disorders exist. showing an overall good impact on self-knowledge about impairments, sense of responsibility and coping skills (e.g. Win, Hassan, Bonney & Iverson, 2015 for a review).

On this topic we found two studies that met inclusion criteria for this revision.

The first study (Thorén et al., 2011) aims to assess the effectiveness of an internet education program guided by an audiologist, comparing it with the effectiveness of an online discussion forum without a professional guidance (control group). It is a pre-post and six months follow-up between group design. Although participants were aged between 24 and 84 years, we chose to include this study in our revision because of mean age of participants was over 60 years with a moderate sloping hearing loss on average. Thorèn et al. (2011) hypothesized that participants to the internet education program would enhance their social participation and communication, increase their hearing aid satisfaction and psychological wellbeing. Moreover, it was hypothesized a maintenance of these positive effects at six months after intervention.

Concerning methodology of intervention, it was thought as a “rehabilitative online educational program” based on a an informational/didactic book “Fading Sounds- About Hearing and Hearing Aids” (Elberling &Worsøe, 2005). In addition, the experimental group received regular email feedback email by an audiologist. Conversely, the control group didn’t get any direct contact with a professional audiologist, but they were asked just to participate at an online discussion group on weekly predetermined topics concerning hearing problems.

Concerning study outcomes, Authors chose four standardized questionnaires in order to assess the reduction in participation restriction and activity limitations, the satisfaction with amplification and level of psychosocial wellbeing at three times: pre, post and at six months follow-up. Reduction in participation restriction and activity limitations was assessed through the HHIE (Ventry & Weinstein, 1982) and it was the primary outcome. Indeed, the first hypothesis was a significant reduction of emotional and social distress (related to restriction and limitation of participation and activities) in participants after the online rehabilitative intervention and at the six-months follow-up when compared to those of control group. Despite this, results from post-hoc analysis showed that a within group effect was found in both groups, with a significant reduction in emotional and social distress both from pre-test to post-test both from pre-test to six months follow up. Nevertheless, not significant between-group effects were revealed by the post-hoc analysis.

Concerning the study outcome on enhancement of psychological wellbeing (assessed through the HADS; Zigmond & Snaith,1983), the hypothesis of this study was that the participants to the online rehabilitative program would have a significant decrease in depression and anxiety scores, compared with the control group and would have maintained it after six months. The results from the study did not fully support this hypothesis: a significant difference in the domain measuring depression after the program was found, but it was not maintained in the six -months follow up. On the other hand, anxiety scores of the study group worsened significantly between pre-test and follow-up. Furthemore, a significant worsening was found in HADS Total Score between post-test and follow-up, whereas no worsening was found in the control group. These results may suggest that the good effects of online rehabilitative intervention as assessed immediately after the program were not maintained after six months.

In a further study, Thorén, Öberg ,Wänström, Andersson and Lunner (2014) extended the previously tested online information-based intervention program, including in it an online peer-to-peer discussion forum and modules derived from the Active Communication Education program (ACE; Hickson, Worral & Scarinci, 2015). Methodology, participants' features, study outcomes and measures were quite similar to the previous research, with two exceptions: in the second study the control group was just referred to a waiting list and time of follow-up was at three months. Summarizing results, the participation to the revised online intervention program significantly affected participants' social and emotional consequences of hearing loss, as assessed by the HHIE, when compared with the control group further on maintaining a significant difference and enhancing improvements at the follow-up.

As regard to the study outcome on enhancement of psychological wellbeing the between-group effects of taking part to the revised online intervention resulted not significant immediately after the program, whilst significant improvements were found in the follow-up, both for depressive both for anxiety symptoms. These results were the opposite of those found in the previous Thorèn et al. (2011) study, when the significant effects assessed immediately after the program, were not maintained in the six months follow-up not for the HHIE neither for the HADS.

Coming from a quite different angle, Kramer and coworkers (2005) aimed to assess the effectiveness of an at home education program for elderly hearing impaired (see Fig. 5.2). Authors hypothesized that taking part to the Home Education Program (HEP), in addition to hearing aid fitting and use, would have positive results in emotional response to hearing loss, in hearing impairment consciousness and communication strategies, also improving interaction with significant others, more than hearing aid fitting alone. In addition, the possibility to follow the program at home was hypothesized to facilitate participation and attendance of older subjects.

Home Education Program, videotapes program's structure (Kramer et al., 2005)	
Film 1 (13 minutes)	"One to one conversation in a quiet room at home"
Film 2 (11,5 minutes)	Birthday party in an noisy environment
Film 3 (14.5 minutes)	Conversation with a stranger
Film 4 (11.5 minutes)	Visit to a doctor in hospital
Film 5 (18 minutes)	Group Meeting with strangers

Fig. 5.2 Outlines of the Home Education Program (Kramer et al., 2005)

Methodologically, 28 hearing impaired adults took part at the study: participants had mean age of 70 years, a moderate hearing loss, all hearing aid users. Subjects were divided into two group: the experimental group that received hearing aid fitting and worked through the 5-12 weeks home educational program (addressed to family members too) and the control group that received HA fitting alone. Concerning reported effects on communication strategies and psychosocial wellbeing, they were assessed by two self-report scale (the Emotional Response scale and the Communication Strategies scale) derived from the Hearing Handicap Disability and Inventory – HHDI (Van den Brink, 1996). Results showed an improvement in communication strategies in the post intervention, maintained in the six-months follow-up as well ($p < 0.05$). These results suggest that participation to the Home Intervention Program increased significantly participants' awareness of communication strategies, such as lipreading and speechreading, also improving interaction with their significant others. Concerning self-perception of quality of life, the administration of IOI (International Outcome Inventory for Alternative Intervention) in the six-months follow-up revealed an improvement in the HEP group, whereas the control group showed a relapse.

Authors (year) Title	**Study design**	**Partecipants**	**Age, yr**	**Better ear pure-tone average† dB HTL**	**Hearing aid fitting %**	**Inclusion criteria**
Andersson, G., et al. (1997) Behavioural hearing tactics: a controlled trial of a short treatment	RCT Experimental between group design pre-post	N = 19 (8 F, 11 M)	Range:67-75 Mean age: 71.5 SD: 2.7	PTA for better ear at 0.5, 1, 2, 4 kHz : 39.7 dB (range 29-52, SD =6.34)	100% hearing aid users Monolateral: 74% Bilateral: 26%	• Hearing impairment • Hearing aids users • Age span: 65-80 • Ability to use telephone • Not previous participation in any rehabilitation course
Kramer, S. et al. (2005) A home educational program for older adults with hearing impairment and their significant others: A randomized trial evaluating short- and long-term effects	RCT	N= 48 (20 F, 28 M) [Exp. group: 24; Control group: 24] n.46 SO [HEP group: 24; Control group: 22]	Range: Not specified [Exp. group Mean age : 69 SD: 7.7; C.group Mean age: 71 SD:8.5	PTA for better ear at 0.5, 1, 2, 4 kHz : [Exp- group: mean 53.7 SD: 13.3; C.group:mean 56.3 SD: 15.7] Moderate typical sloping hearing loss	100% hearing aid users [HEP group Hearing aid users (<1 yrs): 50%: C.group hearing aid users <1 yrs: 37,5%]	• Hearing impairment • Hearing aids users • Age span: over 60 yrs old
Hickson, L. et al. (2007) A randomized Controlled Trial Evaluating the Active Communication Education Program for Older People with hearing impairment	Double-blinded RCT	N=178 (98 F, 80 M)	Mean:73.87 SD:8.29 Range:53–94	Mean:41.33 SD:12.21 Range:13.75–87.50	46% No HA 13% unilateral HA 41% bilateral HA	• Hearing impairment • No significative memory problems or neurological impairment • Attendance at a minimum of three sessions
Thorén, E.S. et al. (2011) Rehabilitative online education versus internet discussion group for hearing aid users: a randomized controlled trial	Randomized control trial Experimental between group design pre-post and 6 mo follow-up	N= 59 (29 w/30m) [I group = 29 C group = 30]	Range: 24-84 yrs Mean age: 63.5 SD: 13.3 yrs	PTA for better ear on average 52 dB HL Moderate typical sloping hearing loss	100% hearing aids users 90% bilateral 10% monolateral	• Hearing impairment • Significative communication difficulties (• At least 1 yr of hearing aid use • Swedish as first language • Access to internet
Saunders, G.H., & Forsline, A. (2012) Hearing aid counseling: Comparison of	Mix method approach	N=74 (7 F,62 M)	Range: 45-75 IC Mean age: 66.1 SD:7.8 PPC mean age: 65.4	PTA – mean of left and right ear on four frequency: Moderate hearing loss	100% bilateral hearing aids	• Hearing imparment • At least 3 months of hearing aid use

single-session informational counseling with single-session performance-perceptual counseling			SD-= 8.6			• Significative dissatisfaction with hearing aids • Age ≥45 years • Not clinical score at the MMSE
Thorén, E.S. et al. (2014) A randomized controlled trial evaluating the effects of online rehabilitative intervention for adult hearing aid users	Randomized control study	N= 76 (32 w/44 m) [I group: N = 38 C group N = 38]	Range: 26-81 yrs Mean age: 69.3 SD: 8.3	PTA for better ear on average 42 dB HL Moderate typical sloping hearing loss	100% ha users 90% bilateral 10% monolateral	• Hearing impairment • Significative communication difficulties (score ≥20 at the HHIE • At least 1 yr of hearing aid use • Swedish as first language • Access to intenet
Oberg, M. et al. (2014)) A preliminary Evaluation of the Active Communication Education Program in a sample of 87-years-old Hearing Impaired Individuals	Within-subject intervention study	23 (11 F, 12 M)	Mean: 87 yrs SD:0	Mean: 48 SD : 8.8	78% HA users 22% No HA users	• Hearing impairment • PTA>25dB HL in at least one ear • Subjective hearing difficulties • Ability to complete at least three sessions Not severe difficulties to communicate in group
Oberg, M., Bohn, T., & Larsson, U (2014) Short- and Long-Term Effects of the Modified Swedish Version of the Actove Communication Education (ACE) Program for Adults with Hearing Loss	Between-group and within-group intervention study	55 (31 F, 24 M)	Mean:69.8 SD:9.7 Range:39-82	Mean:41 SD: 11.8	80% HA users 20% No HA users	• Hearing impairment • PTA>25dB HL in at least one ear • Subjective hearing difficulties • Ability to complete at least three sessions • Not severe difficulties to communicate in group

Table 5.1 Summary table of articles included in final review (RCT = randomised controlled trial, participants reflects number of participants in study)

5.1.1 Considerations on intervention focusing on quality of life in hearing-impaired elderly

On the whole, the presented revision suggests the utility of embracing a holistic vision of auditory rehabilitation in HI elderly, including programs providing clear information, training, and psychosocial support. It is worth pointing out the ecological approach of Kramer et al. (2005) and Hickson et al. (2007) that also included focus on the significant other (spouse, son or whoever important for the hearing-impaired person). The present approach is relevant and seems to be consistent to some extent with the holistic model of handicap and disability (WHO, ICF, 2001). In this regard it has been stressed that hearing disability and impairment need to be read no longer as a feature of the person; rather as an interaction between features of the individual and a number of contextual features (Kramer et al., 2005). As a matter of fact, main aim of Kramer's home educational program was "to raise problem awareness for both the affected individual and the significant other, to enhance communication and to provide knowledge about the nature and consequences of hearing loss" (Kramer et al, 2005, p.256).

Most part of reported studies show a good effectiveness of the rehabilitation programs on level of communication strategy use, activity and participation in old hearing-impaired people.

Nevertheless, the improvement of quality of life and psychological wellbeing after attending programs was not always significant.

Some aspects need a brief observation.

1: Four studies have enhancement of quality of life as their objective. However, we detected a discrepancy under choice of outcome measures. So, Hickson et al. (2007) use the SF-36 (Ware & Shebourne, 1992) that is a health-related quality of life measure, whose items are largely focused on physical functioning and general health. Oberg, Bohn, Larsson and Hickson, (2014) and Oberg, Bohn and Larsson (2014) used only the VAS scale of Euroqol-5d, consisting in just one item ("How good or bad is your health today?"). From a different angle, Saunders and Forsline (2012) used the PIADS -a self-rating scale of psychosocial adjustment (adaptability, self-confidence, and competence)- as outcome measure of quality of life for hearing aids users. We agree that the hearing impaired patient's motivation, the self confidence in their own capacity and competence, the perceived social support, all are crucial elements to front of hearing difficulties and limitations in hearing aid use (Pichora-Fuller, 2016). In any case, the question remains whether consider these dimensions as sufficient measures of quality of life, such as defined according the World Health Organization. As underlined by Boothroyd (2007) "Quality of life reflects self-assessment of the current life experience and includes such things as enjoyment, meaning, purpose, usefulness, value, freedom of choice, and independence. Quality of life is a moving target. It is influenced by function, activity, and participation, but is by no means completely determined by them" (p. 64).

2: Seven studies have enhancement psychological wellbeing and/or psychosocial functioning as a goal of intervention: they evaluate effectiveness of programs or interventions focused on educational enhancement of participation activities and communicative strategies, such as perceived and experienced by elderly hearing impaired subjects. However, in these studies, a quite interchangeable use of term as psychological wellbeing, life satisfaction, psychosocial wellness, is present, often without a clear conceptual framework. Therefore, also the wellbeing measures used in the reported studies seem to assess very different things. Some studies (Oberg, Hickson, 2014; Oberg, Bohn, Larsson, 2014; Thorèn, et al., 2011; Thorèn, et al., 2014) use measures of psychological wellbeing conceptualized as presence/absence of mental or psychological illness. In this regard, psychological wellbeing was just thought as the contrary of “bad-being”. By contrast, Hickson et al. (2007) use a multidimensional measure of psychological wellbeing, conceptualizing it as “positive general functioning” (for example, level of autonomy, personal growth, self-acceptance). Andersson et al. (1997) is the only study we found that focused explicitly on psychological aspects of rehabilitation via teaching techniques of relaxation, coping strategies, hearing tactics and problem-solving procedures. Moreover, it is the only treatment conducted by clinical psychologists. As seen above, the effects on psychological status was assessed by the CSS-CPHI and by the HCA, showing significant treatment effect. Nevertheless, these instruments are not measuring of general psychological or psychosocial wellbeing, just offering an indication of how a subject copes with hearing impairment. With special regard to psychological effects related to hearing loss, we must consider that discrimination, comprehension, and communication difficulties often come on top an age-related psychological framework of increased vulnerability, insecurity, loneliness, and loss of self-confidence. An acquired hearing loss in elderly might deplete psychological functioning and social interactions, increasing feeling of worthlessness, impotence, and isolation. The ability to cope better with hearing loss or to enhance own communicative skills does not mean to gain a better psychological functioning at all.

3: The only study that analyzed the effects of degree of hearing loss on effectiveness of intervention (Oberg, Bohn & Larsson, 2014) showed a variability of benefits in relation to this variable. So, patients with a mild hearing loss improved in communication strategies, whereas patients with a more severe hearing loss- those may have already learned communication strategies- showed better improvement in psychological health.

Considering that the mean degree of hearing loss of the studies' samples was mild/moderate, that might partly justify the scarce effectiveness of reported rehabilitation programs on psychological/psychosocial outcomes.

Moreover, the question arises to what degree weekly group meetings may constitute an obstacle to participation because of a number of logistic problems, concerning both the old patient features both the available professional resources.

In a cost-effective framework the promising results of Saunders & Forsline (2014) study on patient-centered counseling should be considered broadly as an additional tool in routine hearing aid adult old users management. We consider important to underline the noteworthy space given to patients to think about and express their feelings about psychological frustration in daily communication. Nevertheless, a 30 minutes single session counseling might not be enough to benefit from informative counseling content, especially when working with elderly persons. As a matter of fact, Saunders & Forsline (2014) underlined the opportunity to provide at least two counseling sessions including both informations about hearing system and handling of hearing aids both communication strategies and personal concerns.

Clearly there is nowadays a widespread need for more research in order to develop and evaluate effectiveness of additional intervention programs aiming at quality of life and psychological wellness of elderly hearing-impaired population. Starting point of these further research should be the assumption of a circular, ecological and multidisciplinary approach including audiological, medical, functional, social and psychological aspects of the hearing-impaired older adults. As a matter of fact, complementary intervention programs, such as the Active Communication Education program (Hickson et al., 2007) and the Revised Online Education program (Thorén et al., 2014) showed the importance of including both didactic and meta-cognitive procedures, both psychosocial interventions, promoting interaction with peers, sharing of experience and an active problem-solving approach.

5.2 A preliminary evaluation of a Multidisciplinary Communication Rehabilitation Program on a sample of cochlear implant elderly patients

Unfortunately, nowadays, no integrated multidisciplinary rehabilitation programs aiming quality of life and psychological wellbeing in cochlear implant elderly population exist.

The preliminary step of the present project has been to proceed with the translation, and adaptation in Italian language of the Active Communication Education (ACE) program, a communicative rehabilitation program developed at the University of Queensland starting from the research project of Hickson and Worrall (2003). Original version of ACE program was created to help adults with hearing loss to become more effective communicators, providing them with skills and strategies to cope with everyday difficulties. ACE program is intended both for HA users both for nonusers. The ACE handbook defines it as a program "to help adults with hearing loss to become more effective communicators and to provide them with strategies to cope with everyday difficulties" (Hickson, Worral & Scarinci, 2015, p. *V*)

The present “Mind-Active Communication (M-AC) Rehabilitation Program” is intended to be a translation, adaptation, and implementation of ACE program, specifically addressed to cochlear implant elderly users affected by severe-profound hearing loss. It is designed to improve metacognitive awareness, problem-solving and self-management in adults with hearing impairment.

Aims

Primary aim of the present research was to explore the use of an integrated and multidisciplinary rehabilitative program (M-AC) on perceived level of quality of life, psychological wellbeing and hearing abilities self-perception (auditory wellness) in a sample of CI elderly persons aged over 65 years.

Additional aims were to explore the weight of audiological and extra-audiological variables on the outcome of the intervention. and to measure the extent of improvement in perception tests scores and the improvement in verbal recognition.

Audiological variables taken into account were pre-implantation hearing skills, hearing deprivation duration, listening mode (binaural, bimodal, bilateral), years of CI experience. Extra-audiological variables considered were cognitive functioning, level of education, living alone or with significant others.

Materials and Methods

Mind-Active Communication (M-AC) Rehabilitation Program is an integrated program of training group sessions, individual speech therapy and individual psychological counseling, addressing to CI elderly people and encouraging active participation of family members and significant others.

Concerning group sessions, M-AC program is carried out through eight fortnightly sessions lasting about two hours each and for up six to eight participants. Group sessions are conducted by a psychologist, a speech-therapist, and an audiologist: professional figures are intended to have a significant experience in management and care of hearing loss. In each session, presence of family members is highly encouraged.

The aims around which sessions are structured are the expression of communication needs, awareness of obstacles to communication, introduction to a problem-solving approach, and metacognitive control empowerment.

Expression of communication needs: The first two modules introduce the program and include a needs analysis session, during which participants discuss their personal and more significant communication difficulties in their daily use of cochlear implant. Needs and communication demands identified during the first two session will determine the priority of the subsequent addressing issues. An example of schedule meeting topics is shown in Table 5.2. Each session topic is selected starting from communication needs deemed as most important for participants themselves. It is easy to see how M-AC program a not prescriptive, didactic-teaching approach is: contents and topics cannot be strictly precoordinated, as they vary according to the communication difficulties expressed by the participants.

Awareness of communication obstacles: Within each module there is a detailed discussion on the communication activity, on what are the origins of the difficulties, on what the possible solutions, with practical exercises, exercises to do at home and paper information.

Problem-solving approach: The aim is to give participants a series of problem-solving skills (see Figure 5.3) and communication strategies that can be used in a wide range of situations. Between sessions, participants are encouraged to use what they learned during the session in their daily communications. Goal for each participant should be to understand this problem-solving process and then apply it in everyday life. To this end, each activity includes information sheets and practical exercises to do at home and during interval time sessions.

Metacognitive control: it can be defined as the ability to self-evaluate accuracy and adequacy of one's own performance during mental or operational task. It includes self-instructions skills, e.g. to have consciousness of when, how, and why to apply one strategy or another to reach one's own goal. It also includes awareness of one's resources and limits, personal strengths, and weakness (Kluwe, 1982). For example, a person with a good metacognitive control can decide to change strategy in order to get a goal or to go on using the same strategy to solve a problem. M-AC sessions are structured so as to bring the communication activity (characteristics, difficulties and solutions) under the conscious control of the patients through many demonstrations, practical exercises, exchanges between personal and others' experience and observation of alternative behaviors when faced with similar problems.

SESSION	MAIN TOPIC
SESSION 1	PRELIMINARY INFORMATION: COCHLEAR IMPLANT MANAGEMENT AND PRESENTATION OF THE PROGRAM "M-AC"; COMPLETION OF THE COSI QUESTIONNAIRE
SESSION 2	ANALYSIS OF COMMUNICATION NEEDS
SESSION 3	CONVERSATION IN NOISY ENVIRONMENT
SESSION 4	IMPROVE COMMUNICATION SKILLS AND ASSERTIVENESS
SESSION 5	AT HOME COMMUNICATION PROBLEMS
SESSION 6	UNDERSTANDING DIFFICULT SPEAKERS
SESSION 7	USABILITY OF ADVANCED TECHNOLOGY IN ELDERLY
SESSION 8	CONCLUDING TALK AND FINAL REFLECTIONS; COMPLETION OF IOI QUESTIONNAIRE

Table 5.2: Schedule meeting topics of Mind-Active Communication Rehabilitation Program (freely adapted from Hickson, Worral & Scarinci, 2015)

1. What is included in this communication? (who, what, when and why)
2. What is main source of obstacle? (e.g., background noise, too speedy speaker...)
3. What could be a solution? (e.g. Reduction of background noise, to ask to speak slowly…)
4. What information is needed to apply solution? (e.g., knowledge about noise effects on speech comprehension)
5. What skills are needed to apply solutions? (e.g., ability to get help, assertive skills.).

Fig. 5.3 The problem-solving approach (adapted from Hickson, Worral & Scarinci, 2015)

Hearing impaired elderly patients may aslo experience anxiety, depressive mood, low self-efficacy and feeling of worthlessness, risking losing motivation and engagement in cochlear implant use and rehabilitation protocols. So, a further component of M-AC program has been psychological counseling: all participants have benefited from individual counseling sessions provided on fortnightly basis by a clinical psychologist expert in dealing with HI patients. Sessions were held regularly throughout the duration of rehabilitation program, in order to help and support participants long enough to become an “active part”, to become more and more confident and aware of personal strengths and limits, to increase knowledge and metacognitive control.

Concerning sessions of speech therapy, M-AC program includes individual weekly session throughout the 16 weeks program. Sessions are conducted by speech therapists with experience in HL rehabilitation.

Each session was specifically planned in order to:

- get HI elderly fully acquainted with management of hearing devices;
- provide clear information on functioning, cleaning and maintenance of cochlear implant(s) and eventual contralateral hearing aid;

- lead HI old adults to properly use of the aid controls (turn on/ off, change any programs, adjust volume / intensity, enter phone mode, signal pre-processing systems, etc.), to identify and whenever possible resolve major functioning problems.

As for the group training approach, also in individual sessions, a scaffolding approach to metacognitive control is intended as a key element. Special attention is paid to acceptance, understanding and active patient cooperation in finding personal strategies to face with management and care of hearing devices.

Particular attention was paid to the care of listening environment (Pedley & Giles, 2004), identifying and discussing along with the patient which the favorable and which the unfavorable listening situations (for example, proximity to microphones, reduction of background noise, possibility of lip reading, capture attention before starting to speak, adequate speed of speech; presence of background noise, unfavorable conditions of brightness, fast speech, etc.). When necessary, adequate listening assistance systems have been identified (e.g. inductive loops for FM systems, bluetooth interfaces) or visual support (e.g. magnifying glass).

To improve decoding of speech and binaural integration skills, specific listening exercises were performed, with increasing difficulty levels, ranging from auditory attention tasks to speech understanding (Erber, 1982) up to higher levels such as telephone training and perception in noise (Fu & Galvin, 2008). All M-AC program participants was provided with an exercise book, entitled “Hearing Training for Adults with Cochlear Implant” (Nicastri, Mancini, Giallini, Flaccadoro, Amicucci, 2017) (see Fig. 5.4).

Manual intended to offer CI elderly patients an additional in -home training in order to strengthen training and strategies learnt in individual speech-therapist sessions. Material was organized to stimulate different listening skills:

Auditory attention: ability to pay attention to the sounds and noises of one's living environment;

Discrimination: ability to grasp the differences between sound stimuli that can be differentiated by segmental aspects (frequency differences) and by suprasegmental aspects (duration, intensity, intonation, accent);

Identification: ability to identify an auditory stimulus presented within a set of limited alternatives (closed list);

Recognition: ability to identify an auditory stimulus presented with no choice; we usually start by providing help (e.g. semantic or phonological) to get to the actual recognition, where the subject is not given any clue regarding the word / phrase that will be said;

Understanding: ability to interact verbally in communicative situations.

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Materiale per l'allenamento uditivo degli utilizzatori d'impianto cocleare

a cura di: Maria Nicastri, Giulia Taschini, Flavia Flaccadoro, Martina Amicucci

con la collaborazione di: P.Mancini, I. Giallini

Figure 5.4: The Hearing Training Book (Nicastri et al., 2017)

By using the Hearing Training Book (Nicastri et al., 2017), CI elderly patients and significant other (spouse, son, a friend, the therapist) can work on multiple levels simultaneously. Some materials can be used to work on different skills (e.g. identification and recognition of words). Furthermore, the activities can be carried out in easier conditions (proximity to the speaker, absence of environmental noise) or in more difficult ones (in presence of noise, with the speaker at different distances).

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Participants

All participants provided informed consent prior to participation, and all procedures were approved by the local ethics committee of the Cochlear Implant Center. Participants were recruited from Cochlear Implant Center of Organ Sense Department, La Sapienza University of Rome. In total, 43 people aged 65-81 years were identified. All subjects were implanted by two expert oto-surgeons, and currently followed in the Cochlear Implant Center. In all cases, traditional implant surgery has been adopted, characterized by housing the receiver on the temporal scale, mastoidectomy and posterior tympanotomy, cochleostomy at the antero-superior margin of the round window.

Inclusion criteria for the study were: age at the time of study over 65 years; use of unilateral (CI), bilateral (CI / CI) or bimodal (CI / HA) cochlear implants; time of CI experience> 9 months; absence of malformations of the inner ear, ossification / fibrosis of the cochlea and / or incomplete insertions of the array; no significant self-reported history of psychiatric conditions and/or diagnosed incident dementia; normal cognitive level (established as being ≥25° percentile at Raven Coloured Progressive Matrices-CPM (Raven, 1986); attendance at least at 60% of both program group both counseling and speech therapy individual sessions.

Of the 43 eligible subjects, 24 agreed to take part in the research protocol; of those, four subjects did not attend sufficient sessions to be included in the analysis. More specifically, two subjects attended only 30% of sessions because of serious spouse illness; one gave up after two sessions because of cholesteatoma on the implanted ear and one for rupture of the femur. The final number of participants who completed protocol and took part in the study was 20 subjects. Of the 20 participants who attended M-AC program, 9 were male and 11 were women, with a mean age of 72,15 years (65-83; 5,37). Fourteen subjects were married; 4 were widows and 2 were unmarried. Of those unmarried and widows, four lived alone and two lived with family.

Education level was measured by the number of years legally required to attain the education levels that they declared. In the Italian formal education system, compulsory education lasts 8 years; 13 years are needed to obtain a high school diploma and 18 years to reach a college degree. Of the 20 participants, 7 (35%) attended elementary education, 5 (25%) achieved high school diploma and 8 (40%) reached a college degree.

All participants had a CI use experience >11 months; of these, 5 (25%) were CI unilateral, 4 (20%) bilateral and 11 (55%) were bimodal (CI/HA) users. Mean age at cochlear implantation was 67,7 years (59-81; 6,0). Descriptive data of participants are shown in Table 5.3.

All participants were assessed three times: in the pre-treatment selection phase (T0), within one-month post-program (T1) and after six months post-program (T2).

Mean age (range; SD)	72,15 (65-83; 5,37)
Male, n. (%)	9 (45)
Female, n. (%)	11 (45)
Married	14 (70%)
Unmarried	2 (10%)
Widow	4 (20%)
Living alone	4 (20%)
Living with significant others	16 (80%)
Socioeconomic status, n. (%)	
Low	2 (10%)
Medium	16 (80%)
Medium-high	2 (10%)
Smoking, n(%)	
Never	13(65%)
Former	6 (30%)
Current	1 (5%)
Mean age at CI (range; SD)	67,7 (59-81; 6,0)
Mean years of CI experience (range; SD)	4,40 (1-16; 3,89)
Mean years of education (range, SD)	8.95 (5-13; 3,59)
CI bilateral, n. (%)	4 (20%)
CI unilateral, n. (%)	5 (25%)
CI/HA bimodal, n. (%)	11 (55%)

Table 5.3: Descriptive data of the participants (n=20)

Outcome measures

The assessment procedure included audiological, neuropsychological, linguistic, psychological and quality of life aspects, as detailed below.

Only pre-program measures

- Peabody Picture Vocabulary Test (PPVT-R; Dunn & Dunn, 1981; Italian version: Stella, Pizzoli & Tressoldi, 1981): it is a measure of lexical comprehension (receptive auditory vocabulary), with normal standardized scores range between 85 and 115.
- Boston Naming Test (BNT; Kaplan, Goodglass, & Weintraub, 2000; Riva, Nichelli, & Devoti, 2000): the lexical production was measured using the Italian version of this instrument, adapted for school children and normal adults. For this test z scores > 1 are considered normal.
- Raven's Progressive Matrices (colour form): Raven's Progressive Matrices in Colour Form (CPM), developed by John C. Raven in 1947, constitute one of the most used tools for the psychometric measurement of the general components of intelligence or "*g factor*". The Colour Form (CPM) is made up of three series of matrices or figures, for a total of 36 items, which require the solution of visual-spatial problems that involve perceptual-analogical and logical-abstract thought processes. The CPMs have been designed to detect the cognitive level of children, both with typical and atypical development, of adults with mental retardation and of the elderly for detecting deterioration of cognitive abilities. In this context, having regard, in particular to the administration of CPM to the elderly population, comparing the score of the assessed subjects with the scores of the reference sample -reported in the Handbook (Belacchi, Scalisi, Cannoni, & Cornoldi; 2008)- it can be verified how much their current cognitive level falls in the corresponding chronological range, or is higher or lower, showing respectively a better conservation or a decline of intellectual capacity. In our study, the CPMs were used as a selection tool, to verify the absence of significant cognitive impairment, that was an exclusion criterion.
- Repeatable Battery of Assessment of Neuropsychological Status (RBANS; Randolph, Tierney, Mohr, & Chase, 1998): it is an easy-to-use and relatively rapid neuropsychological screening tool (about 40-50 minutes) consisting of twelve tests designed to evaluate five cognitive domains (immediate memory, visuospatial-visuoconstructive skills, language, attention, deferred memory). In Table 5.4 a short description of RBANS domains and their subtests is given.

Domain	Subtest	Description
Immediate Memory	1) List Learning 2) Story Memory	ability to remember informations immediately after their presentation

Visuospatial/constructional ability	3) Figure copy 4) Line orientation	ability to perceive and reproduce dimensional and spatial relations
Language	5) Picture naming 6) Semantic fluency	Ability to respond orally to learned materials that require naming and recall
Attention	7) Digit span 8) Coding (DST)	Ability to remember and manipulate visually and orally information stored in short term memory
Delayed Memory	9) List recall 10) List recognition 11) Story recall 12) Figure recall	Ability to hold for long periods of time (from a few minutes to a lifetime) the information collected

Table 5.4 Description of RBANS domains and subtests

The RBANS, was initially developed as an assessment tool for dementia. It has been validated in “normal” elderly samples (Duff et al., 2003, 2004, 2005) and in some studies on MCI and incident dementia (Juhasz, Kemeny, Linka, Santha, & Bartko, 2003; Kotani et al., 2006). The RBANS has shown adequate sensitivity in detecting cognitive impairment in a number of neuropsychiatric conditions, including Alzheimer's disease (Duff, Hobson, Beglinger, O'Bryant, 2010). The Italian adaptation (Ponteri, Pioli, Padovani & Tunesi, 2007) presents two parallel forms of equal difficulty, such as to allow longitudinal monitoring, reducing the learning effect. The subdomain scores and the total score are age-corrected standard scores, scaled to a normal distribution with a mean of 100 and a standard deviation of 15. Classification of scores is as follows:

>130: Very high performance
120-129: High performance
110-119: In average (high)
90-109: In average (low)
80-89: Below average performance
70-79: Borderline performance
<69: Very low performance

Pre, post and six months post program measures

Assessment of quality of life, depressive mood, psychological wellbeing

- The Hearing Handicap Inventory for the Elderly (HHIE; Ventry & Weinstein, 1982; Ralli, Mizzoni, Clementi & Caramanico, 2014): it is standardized self-report questionnaire, designed to assess the effects of an hearing impairment on emotional and social adaptation in the elderly population. The aim is to identify self-perception of emotional and situational handicap caused by hearing impairment, apart from the objective audiological functioning, considering that correspondence between hearing impairment level and handicap level is not always linear (Ventry &Weinstein, 1982). The tool, widely used as a measure of quality of life in elderly people with hearing loss, consists of 25 questions, for which it is necessary to select one of the three available options ("yes", "sometimes", "no") (see Appendix A). The evaluation of the final score is divided into two categories: emotional sphere (E) and social sphere (S). This will determine a partial score composed of the corresponding scores of each category and a total score that will correspond to the sum of the partial scores (E + S). The tool allows to obtain three results: level of perceived emotional distress, level of perceived social distress and total level of perceived distress related to personal hearing impairment condition.

Scores are expressed as a percentage by dividing the raw score obtained at each subscale by 100. Interpretation of scores is as follows: 0-16% suggest no self-perception of handicap caused by hearing impairment; 18-42% suggest mild-moderate hearing handicap; >44% suggest presence of significant perception of handicap caused by hearing impairment (Newman, Weinstein, Jacobson, Hug, 1991)

- The Psychological General Wellbeing Index (PGWBI) is a questionnaire, widely used internationally as quality of life in clinical trials and epidemiological research (see, e.g., Wiklund I, Karlberg, 1991; Omvik, et al., 1993). More specifically, PGWBI is a measure of the level of subjective psychological wellbeing, assessing self-representation of intrapersonal emotional state and perception of well-being (Grossi & Compare, 2014). PGWBI has been validated and used in many countries, in large populations and in specific groups studies. In 2000 the PGWBI was validated in a representative sample of 1.129 Italian subjects, and its normative values are available (Grossi, Mosconi, Groth, Niero, & Apolone , 2002). The validated Italian version provides a general measure of the self-perceived level of well-being and psychological health. The tool consists of 22 standardized questions, with six alternative answers each, grouped into the following six dimensions of psychological well-being: anxiety, depression, positivity, self-control, general health, and vitality (see Appendix B). Each scale includes 3–5 items. Questions allow multiple choice answers with scores ranging from 0 to 5 (best score value). Depending on studies and authors 'choice, the PGWBI global score varies, between 0 and 110 or between 22 and 132. This bias made comparisons across studies quite difficult. Thus, to facilitate the comparisons of PGWBI scores across studies as well as comparisons with other QoL instruments, for the PGWBI global and partial scores is usually given a normal range of 0 to 100 (Chassany, Dimenas, Dubois,Wu &Dupuy, 2004).

The PGWBI global score represents the sum of all items and can be used as single measure of psychological well-being. and normalized scores range from 0 to 100, with higher scores indicate greater psychological well-being. In order to attribute categorical descriptive properties to the General Well-being index score, the PGWBI Users Manual (Chassany et al., 2004) grouped scores into three broad categories:

66-100 (positive wellbeing)
55-65 (moderate distress)
0-55 (severe distress)

- The Geriatric Depression Scale (Yesavage et al., 1982): it is a self-report questionnaire widely used for the evaluation of depressive symptoms in the elderly, also administered in case of mild or moderate dementia. The GDS consists of 30 standardized items with two alternative questions (yes / no); the tool excludes detection of somatic and psychotic symptoms (see Appendix C). Each answer is assigned a dichotomic 0/1 score; the final score obtained by the subject is categorized as follows: from 0 to 9 (absence of depressive symptoms); from 10 to 19 (presence of mild depressive symptoms); score above 20 (presence of significant depressive symptoms).

Audiological assessment

- Italian Speech Audiometry (Cutugno, Prosser & Turrini, 2000): disyllabic balanced words and sentences, in quiet and in noise (with the speech signal presented at 0° from the participant's head at 65 dB HL and fixed signal-to-noise ratio (SNR +10 e +5) .

- OLSA (Oldenburg Matrix Sentence Test, Italian version; Puglisi et al., 2015): it is based on an adaptive signal-to-noise ratio (SNR) paradigm and it measures the speech reception threshold (SRT) where 50% of low semantically predictable sentences are repeated correctly. The test lists are made up of low semantically unpredictable sentences with a fixed syntactic structure and a random selection of items. Because of the semantically unpredictable structure, the lists cannot be memorized easily and thus, can be used repeatedly.
- STARR Test (Italian version; Dincer D'Alessandro et al., 2016): The STARR test was developed to obtain a reliable SRT assessment with varying signal levels (Boyle et al., 2013). The Italian adaptation (Dincer D'Alessandro et al., 2016) made use of sentences from the standard Italian speech recognition test (Cutugno et al., 2000). The corpus consisted of 10 test lists, each containing 15 sentences, all recorded with a male voice. Three presentation levels (50, 65- and 80-dB HL) were used for sentences, with 5 presentations at each level within a single test list. In the STARR test, 20 dB SNR has been considered to be a cut-off threshold between poor and good users (Dincer D'Alessandro et al., 2018).

For both SRT procedures (Matrix test and STARR test) the participants were recommended to ask for a break whenever needed to avoid excessive strain and performance deterioration.

- The Speech, Spatial and Qualities of Hearing Scale (SSQ) (Gatehouse & Noble, 2004): it is a self-report questionnaire designed to measure a variety of hearing disabilities in a range of different contexts and realistic communication situations. Particular attention is given to the competitive, spatial and movement components of spatial hearing and to the three-dimensional and temporally dynamic aspects of the real auditory world. The SSQ was developed assuming hearing as "scenic analysis": sounds always occur around us virtually, emanating from different sources and several points in time. When a sound is salient, the listener shifts attention, with eyes and head towards the source, and listens carefully: thus, he comprehends sound and can engage in communication and effective dialogue. SSQ consists of three sections:
  - Section one: 14 items on speech hearing: items cover several speech hearing situations, condition of competing sounds, visibility of talkers, number of persons included in conversation and different background conditions.
  - Section two: 17 items on spatial hearing: items include directional and distance judgements, and discrimination of movement.

- Section three:18 items on other hearing qualities: items include ease of listening, naturalness, clarity and identifiability of different speakers, signal segregation, identification/recognition of different musical pieces and instruments, and different everyday sounds.

The questionnaire is preferably to administer in the form of an interview (Noble & Gatehouse, 2004) and participants rated their communication performance in each situation with a score of 0 to 10, with higher scores always reflecting greater ability (or less disability). All subjects were explained that 10 indicated they were able to perform the situation perfectly, whereas 0 indicated they were unable to perform the situation at all. In addition, the option “not applicable” can be checked for cases where the question did not represent an everyday situation.

Statistical analysis

Data are presented as means (standard deviation, sd) or median [range] for continuous variables and when appropriate for categorical variables. The chi-squared test with Yate's continuity correction and Mann-Whitney U test were used to account for differences between continuous variables and proportions, respectively. Analyses were conducted with nonparametric statistics: Friedman test for within-group M-AC comparisons and Wilcoxon pairwise comparisons (pre-program vs. 1-month post-program vs. 6-months post-program) with Bonferroni correction were used to account for the nonparametric distribution.

The relationships between the personal and audiological characteristics of study sample, language skills, and the outcome measures were investigated by the Spearman Rank Correlation Coefficient.

Analyses were carried out using a PC version of Statistical Package for Social Sciences 16.0 (SPSS, Chicago, IL, USA).

Results

Primary outcome: to explore the use and effectiveness of an integrated and multimodal rehabilitative program (M-Ac) on self-perception of social and emotional impact of hearing loss on quality of life, psychological well-being and self-perception of hearing abilities in a sample of cochlear implanted adults aged >65 years.

With respect to the Emotional subscale of HHIE (HHIE-E), in the pre-program assessment (T0), 11 subjects (55% of recipients) had a significant emotional maladjustment to hearing loss, 6 subjects (30%) showed a mild-moderate maladjustment and 3 subjects (15%) revealed a decent emotional adjustment to hearing loss.

After the end of the program (T1) and in the 6-mo follow-up (T2), 11 subjects (55% of recipients) showed mild-moderate emotional maladjustment, 6 (30% of recipients) revealed no hearing-related emotional problems and only 3 subjects (15%) still had severe emotional impairment.

As regard the Social Subscale of HHIE (HHIE-S), at baseline (T0) 14 recipients (70%) showed significant social maladjustment concerning their hearing impairment, 5 subjects (25%) revealed a mild-moderate social suffering and only 1 recipient (5%) showed absence of hearing-related social problems.

At the within 1-mo post-program assessment (T1), the percentage of subjects with severe social problems dropped to 25% (5 recipients); 12 subjects (60%) had mild-moderate social maladjustment and 3 subjects (15%) presented no social suffering related to hearing problems.

At the 6-mo follow-up (T2), the percentage of recipients with severe social problems was unchanged from T1 (5 subjects, 25%), 10 subjects (50%) had mild-moderate hearing social hearing handicap and 5 subjects (25%) reported no hearing handicap social suffering.

At baseline, the observed mean scores for the HHIE were: 45.5 (S.D. 23.5) for Emotional subscale (corresponding to significant handicap); 53 (S.D. 19.6) for Social Subscale (corresponding to significant handicap) and 49.1(S.D. 20.3) for Total Score (corresponding to significant handicap).

After the rehabilitative program (T1), the study group obtained a mean HHIE score of 26.0 (S.D. 16.2) for Emotional subscale (corresponding to mild-moderate handicap); 32.7(14.5) for Social Subscale (mild-moderate handicap) and 29.6(14.4) for Total Subscale.

HHIE scores were not normally distributed (Shapiro–Wilk test, $p<0.05$); nonparametric statistics were used to explore the questionnaire results. Friedman test showed that the change in HHIE emotional, social and total scores across the three measurements was significant (for all three subscales $p < 0.001$).Wilcoxon signed-rank test for within-group M-Ac comparisons (pre-program vs. 1 month post-program vs. 6 mo post-program) showed statistically significant differences in this outcome (HHIE) between measures before (T0) and 1 month after the program (T1) and between before program (T0) and six month follow-up (T2). No statistical differences were found in T1-T2 evaluations.

Pairways comparison with Bonferroni correction pointed out that differences are statistically significant both for T0-T1 emotional subscale (Z=-3,334’ p =.003), both for T0-T1 social subscale (Z=-3,625 , p.= ,000). The rehabilitation program had a positive impact on reducing the social/emotional impacts of hearing loss. The effect size for the comparison is medium, both for emotional both for the social subscale (-0, 53 and -0,57 respectively). Statistically differences were found also in follow-up HHIE measurement (T2), both for emotional (Z=-3,361’ p =.003) both for social subscale (-3,529’ p.= .003), even in this case with a medium effect size (see Table 5.5).

	T0 (n = 20)	**T1 (n=20)**	**P-value T0-T1**	**T2 (n=20)**	**P-value T1-T2**	**P-value T0-T2**
Measures						
HHIE - Emotional	45.5(23.5)	26.0(16.2)	**,003**	23.4(16.38)	,084	**,003**
HHIE - Social	53 (19.6)	32.7(14.5)	**,000**	32.7(17.9)	,512	**,003**
HHIE - Total	49.1(20.3)	29,60(14,4)	**,000**	27,45 (16,19)	,144	**,000**

Table 5.5 HHIE scores and p-value with Bonferroni corrections. Scores are expressed as a percentage by dividing the raw score obtained at each subscale by 100. Interpretation of scores is as follows: 0-16% suggest no self-perception of handicap caused by hearing impairment; 18-42% suggest mild-moderate hearing handicap; >44% suggest presence of significant perception of handicap caused by hearing impairment (Newman, Weinstein, Jacobson, Hug, 1991).

To explore the level of psychological status in our sample of CI elderly person in pre e post-program assessments, the following measures were administered in T0, T1 and in in the 6 months follow up (T2): the GDS to evaluate the presence and intensity of depressive symptoms; the PGWBI to evaluate the level of psychological wellness.

Mean scores and standard deviations at T0, T1 and T2 are presented in Table 5.6.

Concerning the GDS scores, at baseline 9 subjects (45% of recipients) had mild-moderate depressive symptoms, 6 subjects (30%) had no depressive symptoms and 5 subjects (25%) presented significant depressive symptoms.

At the within 1-mo post-program assessment (T1), 15 subjects (75%) showed no presence of depressive symptoms, 4 subjects (20%) had mild-moderate symptomatology and only one recipient still had significant depressive symptoms (5%).

At the 6-mo follow-up (T2), 13 subjects (65%) showed no depressive symptoms and 7 (35%) had a mild-moderate depressive symptomatology. No recipients (0%) revealed presence of significant depressive symptoms.

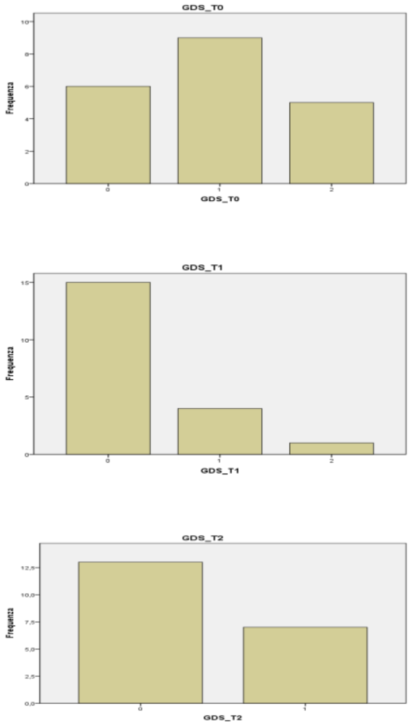

Figure 5.4. Geriatric Depression Scale: graphical informations of the frequencies of depressive symptoms at T0, T1 and T2

Wilcoxon signed-rank test for within-group M-Ac comparisons (pre-program vs. 1 month post-program vs. 6 months post-program) showed statistically significant differences between measures before (T0) and 1 month after the program (T1) both for GDS (z= -3,656, p.=.000) both for General Wellbeing (PGWBI Total score; z=-2,923, p-=.003). Concerning single domains of PGWBI, statistically significant difference in T0-T1 assessments were found for the following subscale: anxiety (-2,666, p.=.008), depression (-2,311, p.=.021), positivity (2,173, p.=.021), and vitality (z= -3,178, p-=.030). A positive trend was found also in self-control subscale, but not significant at the 95% significance level (z= -1,938, p-=.053). Statistically differences were found also in follow-up T2 GDS and in PGWBI subscales of Anxiety, Depressive mood, Positivity and Vitality (see Table 5.6).

	T0 (n = 20)	**T1 (n=20)**	**P-value T0-T1**	**T2 (n=20)**	**P-value T1-T2**	**P-value T0-T2**
Measures						
Geriatric Depression Scale	12.5 (5.47)	8.5 (4.76)	**,000**	8.3(3.77)	,72	**,000**
PGWBI (Total)	65,20 (14,4)	72,40 (12,88)	**,003**	73,35 (13,8)	,977	**,014**
Anxiety	65,80 (18,82)	75,80 (15,92)	**,008**	74,55 (18,2)	,975	**,007**
Depressive mood	76,30 (16,41)	83,40 (13,65)	**,021**	83,60 (13,6)	,782	**,038**
Positivity	53,75 17,462	60,40 (17,7)	**,030**	63,40 (19,4)	,129	**,009**
Self-control	73,35 17,614	79,80 (18,2)	,053	78,50 (16.3)	,593	,142
General health	65,20 17,978	72,40 (20,1)	,233	73,35 (21,6)	,598	,678
Vitality	63,15 20,597	71,25 (18,9)	**,001**	71,75 (18,3)	,887	**,003**

Table 5.6 GDS and PGWBI mean scores and standard deviations at T0, T1 and T2

To assess subjective experience and quantify listening disabilities in realistic communication situations (auditory wellness), the SSQ was administered three times (T0; T1;T2).

Table 5.7 shows mean scores (and SDs) in T0, T1 and T2 on each section in the SSQ, namely Speech Hearing, Spatial Hearing and Quality of Hearing sections.

As SSQ scores were not normally distributed (Shapiro–Wilk test,$p<0.05$); nonparametric statistics were used to explore the questionnaire results. Wilcoxon signed-rank test for within-group M-Ac comparisons (pre-program vs. 1-month post-program vs. 6-months post-program) showed statistically significant differences in SSQ scores between measures before (T0) and 1 month after the program (T1). Differences are statistically significant both for speech hearing subscale (z =-3,884 , p =.000), both for spatial hearing subscale (-3,921, p.= .000), both for quality of hearing subscale (-3,921, p.= .000)

SSQ Section	**T0 (n = 20)**	**T1 (n=20)**	**P-value T0-T1**	**T2 (n=20)**	**P-value T1-T2**	**P-value T0-T2**
Speech hearing items	2,9 (1,6)	4,6 (1,3)	**.000**	4,8(1,9)	,507	**.006**
Spatial hearing items	3,1 (1,8)	4,9 (1,4)	**.000**	4,7 (2,1)	,530	,079
Quality of hearing items	3,8 (1,7)	5,8 (1,6)	**.000**	6,1 (1,9)	,814	**.001**

Table 5.7 SSQ mean scores and standard deviations at T0, T1 and T2

In pre-program assessment (T0) mean scores of speech hearing section items were slightly lower (greater disability) than the other sections, with highest mean scores provided by quality of hearing subscale. The highest ratings for quality of hearing section continued to be stable across assessments in T1 and T2 with significant improvement in mean scores (lower disability) across all SSQ sections. In follow-up, quality of hearing scores showed the highest significant improvement compared to pre-program assessment (-3,295, p.= .001) (see Table 5.7).

Further aims of this work were to define the weight of audiological and extra-audiological variables on the primary outcome (HHIE) of the intervention and to measure the extent of improvement in perception tests scores and the improvement in verbal recognition.

Of all subjective variables that may affect emotional and social distress related to hearing impairment, the gender, the socioeconomic status, the marital status, living alone or with significant others, the educational level, the general neuropsychological functioning and the age of participants did not affect the HHIE scores in any of the three measurements (T0,T1,T2) (all p values > 0.1). Concerning neuropsychological functioning, in Table 5.8 the mean RBANS total score and subscale scores are presented. Evaluation was carried out on all participants before the beginning of MA-C program, to get a neuropsychological screening of the sample.

RBANS Domain	**Mean Score (sd)**	Description
Immediate memory	95.1 (sd. 17.2)	In average
Visuospatial ability	95.9 (sd 12.2)	In average
Language	88.4 (sd 9)	Below average
Attention	86.7 (sd 16.2)	Below average
Delayed Memory	98.7 (sd 13.5)	In average
General Score	88.2 (sd. 12.03)	Below average

Table 5.8 Mean e standard deviation RBANS scores in pre-program assessment

The scores from RBANS in our sample are slightly lower than findings from Claes et al. (2018) study on cognitive Performance of CI older adults. Anyway, in Claes et al. study (2018), authors used an alternative version of RBANS (RBANS-H), especially developed to examine cognition in individuals with a hearing impairment and evaluation was made preimplantation and at 6 and 12 months after implantation. In our sample, the years of cochlear implant experience vary from less than 1 years up to 16 years; moreover in the alternative version used by Claes et al. (2018) stimuli are presented in audio-visual way, probably making the tasks simpler for CI subjects.

For audiological variables, years of CI experience, type, side of implantation (monolateral, bilateral or bimodal) and coding strategy did not affect any of HHIE outcomes.

By contrast, duration of hearing loss seemed to affect HHIE scores for the emotional subscale in post program (T1) and follow-up assessments (T2) (all p-values<0.01). Recipients whose hearing impairment had arisen earlier, showed in T1 and T2 a lower percentage of better scores than subjects with a more recent hearing loss.

This finding is partially consistent with Oberg et al. (2014) study. In fact, it is possible that people with a longer experience of hearing loss may have built long-term strategies to cope with hearing difficulties. These strategies may have now become too rigid to respond positively to a rehabilitation approach, although offering alternative or more functional coping skills. Moreover, referencing to Horn and Cattell (1967) and Salthouse (1994), a crystallization of these long-term skills may have occurred with a subsequent resistance to change and modification. Hence, when a long-term hearing- impaired old person is asked to process unfamiliar stimuli or new task involving metacognitive and executive engagement, performance may be depleted.

A significant correlation was found between a higher presence of depressive symptoms at baseline (as measured by GDS) and worse level of perceived emotional and social distress in T0 (all p values <0.01). We found also significant correlation in T1 between a higher presence of depressive symptoms and worse scores in emotional distress (p =.028) and in T2 between in higher depressive symptomatology and both emotional and social distress ((all p values <0.05).

For psychological wellbeing dimensions, with exception an Anxiety scores, for all dimensions of PGWBI a significant correlation with HHIE scores both in emotional and social scale was found in pre-program assessments (all p-values<0.05). By contrast no significant correlation between PGWBI scores and HHIE outcomes was found in T1 and T2 assessments.

Post-rehabilitation evaluation showed significant improvement in all verbal perception tests, even in a complex task such as consonant confusion (pre / post = 47/58%; pre / post sentences = 78/92%; p <0.05). After rehabilitation, a greater tolerance to sound with cochlear implant and a significant improvement in the threshold in the free field (pre / post = 34 / 28.4 dB; p <0.05) has been found. The formal tests in noise showed an improvement in the recognition of words and phrases in both quiet and fixed noise and in adaptive noise: words SNR + 10 pre / post = 32/46%; STARR pre / post test = 18.8 / 11.9 dB SNR (p <0.05).

Verbal perception of consonant and disyllabic confusion and recognition in noise were statistically correlated (Spearman's Ro) to the reduction of emotional and social difficulties reported by the elderly in the HHIE questionnaire (p <0.05). In addition, the elderly report a significant increase in the quality of speech understanding (pre / post = 3.8 / 5.09), in the ability to localize sounds in everyday life (pre / post = 4 / 5.5) and in quality of sounds and voices heard through the cochlear implant (pre / post = 4.25 / 6.25) (p <0.01).

Discussion and preliminary conclusions

This pilot study was a first attempt to evaluate use and effectiveness of a multidisciplinary “scaffolding” approach to rehabilitation of CI elderly.

The importance of including meta-cognitive procedures, and psychological/counseling interventions in rehabilitation of hearing-impaired elderly has been emerging for a few years now, as evidenced by the literature minireview reported in the introduction of this chapter. The lack of this type of intervention aimed at the CI elderly persons prompted us to undertake the presented project

A first indication that the program might be beneficial is a significant improvement in the level of hearing-related emotional and social adaptation (as measured by HHIE) and in psychological wellbeing (as measured by GDS and PGWBI) after participation in the rehabilitation protocol. Moreover, these improvements were observed also in the six months follow-up, both for HHIE, both for level of anxiety, depressive mood, positivity, and vitality. These results are consistent with Oberg, Bohn & Larsson (2014); anyway, it is difficult to compare the two studies because of the use of different questionnaires and the different nature of the participant samples.

It should be emphasized that HHIE is a self-assessment tool for quantification of the emotional and social effects of self-perceived hearing loss in the elderly. We used it as primary outcome measure to assess the level of perceived hearing -related quality of life in a sample of CI old adults. Anyway, the concept of quality of life is broader than health-related problems. As underlined by Boothroyd (2007) “Quality of life reflects self-assessment of the current life experience and includes such things as enjoyment, meaning, purpose, usefulness, value, freedom of choice, and independence. Quality of life is a moving target. It is influenced by function, activity, and participation, but is by no means completely determined by them” (p. 64). So, in the present study, additional instruments have been used in order to obtain a more complete evaluation of quality of life outcomes, including the Geriatric Depression Scale and the Psychological General Well-Being Index, as measures of the level of subjective psychological wellbeing, self-representation of intrapersonal emotional state and perception of well-being (Grossi & Compare, 2014).

Concerning emotional and social adjustment to hearing and communicative problems, functionalist theories on emotional regulation emphasize how emotions can facilitate adaptation to the environment, leading to an easier decision making, better cognitive and attentional processes, recording significant events in memory, also providing information regarding the correspondence between the organism and the surrounding environment (Schwartz & Clore, 2003). So, for example, the “Feeling-as-Information Theory” (Schwartz, 2010) states that subjective experiences –moods, emotions, metacognitive experiences, and bodily sensations – have a crucial role in personal judgements of social situations. “It assumes that people attend to their feelings as a source of information, with different feelings providing different types of information" (Schwartz, 2010, p. 290) Modern research on the psychology of emotion regulation underlined the existence of adaptive and maladaptive strategies that can be used in emotion regulation. The emotion-generating systems that are targeted in emotion regulation include attention, knowledge, and bodily responses. Reappraisal, problem solving strategies, self-acceptance are considered adaptive strategies for emotion regulation. Reappraisal consists in the generation of positive interpretations or perspectives on a stressful situation, in order to reduce its negative effects; problem-Solving is the conscious attempt to change a stressful situation or to contain its consequences. Finally, acceptance can be defined as the non-judgmental acceptance of his own emotional experience.

Both the group training both the individual sessions of M-Ac rehabilitation base on a scaffolding approach to metacognitive control, with special attention to acceptance, understanding and active patient cooperation in finding personal strategies to face with management and care of hearing devices. Anyway, recipients whose hearing impairment had arisen earlier, showed in T1 and T2 a lower percentage of better scores than subjects with a more recent hearing loss. This finding is partially consistent with Oberg et al. (2014) study. In fact, it is possible that people with a longer experience of hearing loss may have built long-term strategies to cope with hearing difficulties. These strategies may have now become too rigid to respond positively to a rehabilitation approach, although offering alternative or more functional coping skills. Moreover, referencing to Horn and Cattell (1967) and Salthouse (1994), a crystallization of these long-term skills may have occurred with a subsequent resistance to change and modification. Hence, when a long-term hearing-impaired old person is asked to process unfamiliar stimuli or new task involving metacognitive and executive engagement, performance may be depleted. Moreover, we also consider that the lack of an effective communication determines more and more interactive breakdown with an increasing sense of discomfort and inability to share a rich and informative communication. When communicative breakdowns occur again and again, an aged person often cares to be considered an "old dotard", because of the overlapping effects of hearing loss and the presence of age-related "slow- down" in cognitive processes. As Vermeire, Brokx, Wuyts, Cochet, Hofkens, & Van de Heyning (2005) underlined, foundation of self-reliance and sense of independence in elderly is even related to self-perceived communication abilities and self-control.

A further component of M-AC program has been psychological counseling, addressed to promote well-being and optimize elderly resources.

In psychological counselling to hearing impaired elderly, it it is useful to pay attention to some methodological measures. There are a lot of evidence that the lower and lower motivation to communicate may finally impede participation in social life, relational, cultural and aggregation activities (e.g. Kramer, 2005; Heine & Browning, 2002; Boothroyd, 2007; Hickson et al., 2006; 2007a; 2007b; Covelli et al., 2015; Lin et al., 2012): this, it is necessary to consider the emotional consequences of a daily experience of reduced speech perception and comprehension and the consequences of constant hampered and depleted communication. Moreover, in addition to hearing difficulties, age-related attention, memory, visual problems need to be taken into account. So, it is helpful to compensate for it with concrete examples, suggestions, repetition. We consider flexibility and attunement two key factors in psychological support to elderly, especially when faced with a severe sensory deprivation. So, founding elements of psychological approach to participants have been active listening, empathic participation, emotional availability, plain language, eye contact, friendly gestures, in order to communicate acceptance, authenticity and empathic understanding (Rogers, 1978). The final aim was to offer an emotional scaffolding for more successful clinical and rehabilitative outcomes. As a matter of fact, the concept of "actualizing tendency", largely used by Rogers' approach to patients, refers to the intrinsic ability in the human being to selectively and directly orient himself towards the completion and actualization of his potential.

Thus, the decrease in self-perception of emotional and situational distress and the enhancement in psychological general well-being can be also explained as a consequence of an increased awareness of the predictability of some challenging social situations, resulting in a decrease in the levels of anxiety, insecurity and social tension (Andersonn et al., 1997) and in a better use of the personal resources. As a matter of fact, increasing motivations and a positive emotional attitude allow people to make the best use of the cognitive resources preserved (Cornoldi, 2011). Knowledge and metacognitive control are not distinct elements, on the contrary, control is absolutely tied to the knowledge possessed, as well as knowledge find strength from previous experiences and how these experiences have been addressed, resolved and understood, also enhancing sense of self-efficacy (Coutinho, 2008) and the chance to invest in one's self.

So, the significant improvements in psychological wellbeing after participation at the program might indicate that a mixture of approaches leading to a larger comprehension of audiological and hearing dimensions, acquisition of communicative and pragmatic strategies to front of daily conversation obstacle and, in addition, a problem-solving and interactive methodology can generate a general growing of self-confidence and reliance on own skills, giving rise to positive feelings of self-acceptance, self- responsibility and assertiveness, finally improving quality of life of CI elderly patients.

The presence of significant others to the M-Ac group session and their active and constant involvement in individual speech-therapy sessions is another element to consider. It is possible that an increased awareness of the experiences of coping with the multiple hurdles along the way of hearing impairment and rehabilitation, have had a positive impact on outcomes. Studies on this topic showed mixed results (Preminger and Meeks, 2010; Hickson et al., 2007; Oberg et al., 2014). Anyway, further investigation on this topic are needed.

No significant correlation emerged between the improvement in vitality and well-being scores and scores on the verbal perception tests of consonant and disyllabic confusion and recognition in noise. However, the post-rehabilitation evaluation shows a significant decrease in depressive symptoms, assessed through the GDS, which notes that, at the end of the MA-C program, none of the participants had severe depressive symptoms. This improvement, not related to the auditory aspects, is probably linked to the acquisition of the strategies for the prevention and management of communication difficulties presented and practiced during the rehabilitation process and to the level of depression and anxiety.

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This was a first attempt to evaluate the use and effectiveness of the Mind-Active Communication (M-AC) Rehabilitation Program. It is an integrated program of training group sessions, individual speech therapy and individual psychological counseling, Further studies, including a large sample and a control group are needed. Anyway, this final consideration can be made: rehabilitative approach aiming to multilevel skills, such as, comprehension of audiological and hearing dimensions, acquisition of communicative, pragmatic and problem-solving strategies, implements of interaction and sharing of experience with peers, together with a psychological counselling might help CI elderly in growing self-confidence and reliance on own skills, giving rise to positive feelings of self-acceptance, self- responsibility and assertiveness. In turn, an improvement of these aspects can significantly promote optimal use of the cochlear implant even in the elderly, reducing the risk of losing motivation and engagement in cochlear implant use and rehabilitation protocols.

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Conflict of interest

The author(s) declare that they have no conflict of interest.

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Appendix A: *Questionario di screening uditivo: Hearing Handicap Inventory for the Elderly*

s1. Un problema di udito ti obbliga a usare il telefono meno di quello che ti piacerebbe fare?	**No**	**Qualche volta**	**Si**
e2. Un problema di udito ti crea imbarazzo quando conosci nuove persone?	**No**	**Qualche volta**	**Si**
s3. Un problema di udito ti costringe ad evitare la compagnia di altre persone?	**No**	**Qualche volta**	**Si**
e4. Un problema di udito ti rende irritabile?	**No**	**Qualche volta**	**Si**
e5. Un problema di udito ti fa sentire frustrato mentre parli con i tuoi famigliari?	**No**	**Qualche volta**	**Si**
s6. Un problema di udito ti crea difficoltà a partecipare ad una festa?	**No**	**Qualche volta**	**Si**
e7. Un problema di udito rende difficile ascoltare e capire i colleghi, i collaboratori, i clienti?	**No**	**Qualche volta**	**Si**
s8. Ti senti handicappato a causa del problema di udito?	**No**	**Qualche volta**	**Si**
e9. Un problema di udito ti fa sentire frustrato quando ti trovi con gli amici, i parenti, i vicini?	**No**	**Qualche volta**	**Si**
s10. Un problema di udito ti fa sentire frustrato quando parli con colleghi, collaboratori, clienti?	**No**	**Qualche volta**	**Si**
s11. Un problema di udito ti crea problemi al cinema e/o a teatro?	**No**	**Qualche volta**	**Si**
e12. Un problema di udito ti rende nervoso?	**No**	**Qualche volta**	**Si**
s13. Un problema di udito ti costringe a fare meno visite agli amici, ai parenti, ai vicini rispetto a quanto vorresti?	**No**	**Qualche volta**	**Si**
e14 . Un problema di udito causa delle discussioni in famiglia?	**No**	**Qualche volta**	**Si**
s15. Un problema di udito ti causa problemi quando ascolti la radio o la televisione?	**No**	**Qualche volta**	**Si**
s16. Un problema di udito ti costringe a visitare meno i negozi di quanto vorresti?	**No**	**Qualche volta**	**Si**
e17. Un qualsiasi problema o difficoltà nell'udito ti sconvolge completamente?	**No**	**Qualche volta**	**Si**
e18. Un problema di udito ti costringe a restare da solo/a?	**No**	**Qualche volta**	**Si**
s19. Un problema di udito ti obbliga a parlare meno con i famigliari rispetto a quanto vorresti?	**No**	**Qualche volta**	**Si**
e20. Ti sembra che qualsiasi difficoltà con il tuo udito limiti od ostacoli la tua vita personale e sociale?	**No**	**Qualche volta**	**Si**
s21. Un problema di udito ti crea difficoltà quando ti trovi in un ristorante con amici o parenti?	**No**	**Qualche volta**	**Si**
e22. Un problema di udito ti fa sentire depresso?	**No**	**Qualche volta**	**Si**
s23. Un problema di udito ti obbliga ad ascoltare meno radio e tv di quello che vorresti?	**No**	**Qualche volta**	**Si**
e24. Un problema di udito ti fa sentire a disagio quando parli con gli amici?	**No**	**Qualche volta**	**Si**
e25. Un problema di udito ti fa sentire escluso quando ti trovi in un gruppo di persone?	**No**	**Qualche volta**	**Si**

Appendix B: *Psychological General Well-Being Index*

QUESTIONARIO PER LA VALUTAZIONE DELLO STATO GENERALE DI BENESSERE PSICOLOGICO

The Psychological General Well-Being Index (PGWBI)
Dupuy H.J., 1984; Versione MiOS, Gennaio 2000
Questo questionario si propone di verificare il Suo stato attuale di benessere ponendoLe alcune domande su "come si sente" e su come Le stanno andando le cose in generale.
Dopo aver letto attentamente tutte le possibili risposte, scelga per ciascuna domanda la risposta che Le sembra descrivere meglio la Sua situazione.

1. Nelle ultime 4 settimane, come si è sentito in generale?
2. Nelle ultime 4 settimane, è stato infastidito da malattie, disturbi fisici o dolori?
3. Nelle ultime 4 settimane, si è sentito depresso?
4. Nelle ultime 4 settimane, si è sentito padrone delle Sue situazioni, pensieri, emozioni e dei Suoi sentimenti?
5. Nelle ultime 4 settimane, è stato infastidito da stati di tensione o perché aveva i nervi a fior di pelle?
6. Nelle ultime 4 settimane, quanta energia o vitalità ha avuto o ha sentito di avere?
7. Nelle ultime 4 settimane, mi sono sentito scoraggiato e triste.
8. Nelle ultime 4 settimane, è stato generalmente teso o ha provato tensione?
9. Nelle ultime 4 settimane, in che misura si è sentito felice, soddisfatto o contento della Sua vita personale?
10. Nelle ultime 4 settimane, si è sentito così bene da fare quello che desiderava o doveva fare?
11. Nelle ultime 4 settimane, si è sentito tanto triste, scoraggiato, disperato o ha avuto tanti problemi da chiedersi se valesse la pena andare avanti?
12. Nelle ultime 4 settimane, mi sono svegliato fresco e riposato.
13. Nelle ultime 4 settimane, ha provato apprensione, preoccupazione o paura per la Sua salute?
14. Nelle ultime 4 settimane, ha avuto qualche motivo per domandarsi se stesse perdendo la ragione o se stesse perdendo il controllo della memoria, dal modo in cui agisce, parla, pensa o sente?
15. Nelle ultime 4 settimane, la mia vita quotidiana è stata interessante per me.
16. Nelle ultime 4 settimane, si è sentito attivo, in forze o lento, pigro?
17. Nelle ultime 4 settimane, è stato in ansia, preoccupato o arrabbiato?
18. Nelle ultime 4 settimane, mi sono sentito emotivamente stabile e sicuro di me stesso.
19. Nelle ultime 4 settimane, si è sentito rilassato, tranquillo oppure si è sentito molto teso, nervoso o agitato?
20. Nelle ultime 4 settimane, mi sono sentito allegro e sereno.
21. Nelle ultime 4 settimane, mi sono sentito stanco, esaurito, logorato o sfinito.
22. Nelle ultime 4 settimane, è stato o si è sentito sottoposto a stress o pressioni?

Appendix C: *Geriatric Depression Scale*

(Yesavage JA, Rose TL, Lum O, Huang V, et al. Development and validation of geriatric depression screening: a preliminary report. J Psychiatr Res 1983;17:37-49)

1 E’ soddisfatto della sua vita?

2 Ha abbondonato molte delle sue attività e dei suoi interessi?

3 Ritiene che la sua vita sia vuota?

4 Si annoia spesso?

5 Ha speranza nel futuro?

6 E’ tormentato da pensieri che non riesce a togliersi dalla testa?

7 E’ di buon unore per la maggior parte del tempo?

8 Teme che le stia per capitare qualcosa di brutto?

9 Si sente felice per la maggior parte del tempo?

10 Si sente spesso indifeso?

11 Le capita spesso di essere irrequieto e nervoso?

12 Preferisce stare a casa, piuttosto che uscire a fare cose nuove?

13 Si preoccupa frequentemente per il futuro?

14 Pensa di avere più problemi di memoria della maggior parte delle persone?

15 Pensa che sia bello stare al mondo, adesso?

16 Si sente spesso abbattuto e triste adesso?

17 Trova che la sua condizione attuale sia indegna di essere vissuta?

18 Si tormenta molto pensando al passato?

19 Trova che la vita sia molto eccitante?

20 Le risulta difficile iniziare ad occuparsi di nuovi progetti?

21 Si sente pieno di energia?

22 Pensa di essere in una situazione priva di speranza?

23 Pensa che la maggior parte delle persona sia in una condizione migliore della sua?

24 Le capita spesso di turbarsi per cose poco importanti?

25 Ha frequentemente voglia di piangere?

26 Ha difficoltà a concentrasi?

27 Si alza con piacere la mattina?

28 Preferisce evitare gli incontri sociali?

29 Le riesce facile prendere delle decisioni?

30 Ha la mente lucida come prima?

Punteggio totale ____/30