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Long-term speech perception and morphosyntactic outcomes in adolescents and young adults implanted in childhood

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Abstract:	Background: Long-term assessments of children with cochlear implants (CI) are important inputs to help guide families and professionals in therapeutic and counselling processes. Based on these premises, the primary aim of the present study was to assess the long-term speech and language outcomes in a sample of prelingually deaf or hard of hearing (DHH) adolescents and young adults with unilateral or bilateral implantation in childhood. The secondary aim was to investigate the correlations of age at implantation with long-term speech and language outcomes. Materials and Methods: Retrospective observational study on 54 long-term CI users, 33 unilateral and 21 bilateral (mean age at CI surgery 38.1 ± 24.6 months; mean age at last follow-up assessment 19.1 ± 4.3 years of age and mean follow-up time 16 ± 3.7 years). Means and standards were used to describe speech perception (in quiet, in fixed noise and in adaptive noise using It-Matrix) and morphosyntactic comprehension (TROG-2) outcomes. A univariate analysis was used to evaluate outcome differences between unilateral and bilateral patients. Bivariate analysis was performed to investigate the relationships between age at CI, audiological variables, and language outcomes. Finally, multivariate analysis was performed to quantify the relationship between It-Matrix, sentence recognition in quiet and at SNR+10 and TROG-2. Results: The participants showed good speech recognition performance in quiet (94% for words and 89% for sentences) whilst their speech-in-noise scores decreased significantly. For the It-Matrix, only 9.2% of the participants showed scores within the normative range. This value was 60% for TROG-2 performance. For both auditory and language skills, group differences for unilateral versus bilateral CI users were not statistically significant ($p > 0.05$). Bivariate analysis showed that age at CI correlated significantly with overall results at TROG-2 ($r = -0.6$; $p < 0.001$) and with It-Matrix ($r = 0.5$; $p < 0.001$). TROG-2 was negatively correlated with results for It-Matrix ($r = -0.5$; $p < 0.001$). In the multivariate analysis with It-Matrix as a dependent variable, the model explained 63% of the variance, of which 60% was related to sentence recognition and 3% to morphosyntax. Conclusions: These data contribute to the definition of average long-term outcomes expected in subjects implanted during childhood whilst increasing our knowledge of the

	effects of variables such as age at CI and morphosyntactic comprehension on speech perception. Although the majority of this prelingually DHH cohort did not achieve scores within a normative range, remarkably better It-Matrix scores were observed when compared to those from postlingually deafened adult CI users.
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SAPIENZA
UNIVERSITÀ DI ROMA

Rome, 10 March 2023

To **Joseph E. Kerschner**

Editor-in-Chief of International Journal of Pediatric Otorhinolaryngology

Dear Editor,

On behalf of all authors, please find attached the revision of our manuscript entitled "**Long-term speech perception and morphosyntactic outcomes in adolescents and young adults implanted in childhood**" for consideration by the International Journal of Pediatric Otorhinolaryngology.

As one of the primary authors and on behalf of all my coauthors, I declare that:

1. All of the authors listed in the byline have made contributions appropriate for assumption of authorship, have consented to the byline order, and have agreed to submission of the manuscript in its current form. As the primary author I ensure that all my co-authors have played significant roles in writing the manuscript, designing the study, preparing and executing the plan for data collection, and interpreting the results in preparation for publication.
2. This retrospective clinical research adheres to basic ethical considerations for the protection of human participants in research and the paper has been accepted by the Ethical Institutional Review Board of "Policlinico Umberto I" of Rome-Italy.
3. The present manuscript has not been previously published in the same, or essentially the same, form, and it is not currently under review elsewhere.
4. The Authors declare NO real or potential conflicts of interest that could be seen as having an influence on the research (e.g., financial interests in a test or procedure, funding by an equipment or materials manufacturer for efficacy research).

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Thank you for your consideration of this manuscript.

Sincerely,

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To the editor and reviewers:

We thank the reviewer for the comments. Please find enclosed a point-by-point answer. The modifications in the text were marked in red.

Rephrase the first sentence of the results section in the Abstract, condensing perceptual and grammatical outcomes.	Thanks for the suggestion, the results section of the abstract has been rephrased accordingly.
In the conclusions of the abstract and of the paper please report that 60% of the pts had grammatical scores within normal limits	Thank you for the suggestion. The conclusion section of the abstract has been revised accordingly.
The references do not report the pages of the papers	According to the IJPORL style, Mendeley reference management software was used. Page numbers and DOI are now added to each reference.
Line 38 It would be preferable to define what "hard of hearing (DHH) children" means.	Line 38 has been modified accordingly. A reference was added.
Line 85-109 This part should be included in the "discussion"	As suggested, most of the content of this section has been moved to the discussion section thereby reducing the length of the introduction.
The introduction is very long and redundant, it is advisable to summarize the basic concepts of the literature and focus on the purposes of the study.	Thank you for the suggestion. The Introduction has now been revised accordingly.
Line 146 "for unilateral users the average pure tone audiometry (PTA) threshold for residual frequencies was 110.2 dB HL" it means the threshold of the contralateral ear? The unilateral users used hearing aid in the contralateral ear. This aspect should be clarified	Following the modification to the abstract and introduction this sentence is now in line 117-119. The manuscript has been improved by a better explanation of these aspects.
Line 356-368 this part as other parts that describe literature overview "weigh down" the paper…I would consider turning them into tables or summarize them.	Following this suggestion, we have shortened this part of the Discussion whilst keeping some important outcome comparisons to help facilitate a better understanding of the present findings.

LONG-TERM SPEECH PERCEPTION AND MORPHOSYNTACTIC OUTCOMES IN ADOLESCENTS AND YOUNG ADULTS IMPLANTED IN CHILDHOOD

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Declaration of interests

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Long-term speech perception and morphosyntactic outcomes in adolescents and young adults implanted in childhood

Abstract

Background: Long-term assessments of children with cochlear implants (CI) are important inputs to help guide families and professionals in therapeutic and counselling processes. Based on these premises, the primary aim of the present study was to assess the long-term speech and language outcomes in a sample of prelingually deaf or hard of hearing (DHH) adolescents and young adults with unilateral or bilateral implantation in childhood. The secondary aim was to investigate the correlations of age at implantation with long-term speech and language outcomes.

Materials and Methods: Retrospective observational study on 54 long-term CI users, 33 unilateral and 21 bilateral (mean age at CI surgery 38.1 ± 24.6 months; mean age at last follow-up assessment 19.1 ± 4.3 years of age and mean follow-up time 16 ± 3.7 years). Means and standards were used to describe speech perception (in quiet, in fixed noise and in adaptive noise using It-Matrix) and morphosyntactic comprehension (TROG-2) outcomes. A univariate analysis was used to evaluate outcome differences between unilateral and bilateral patients. Bivariate analysis was performed to investigate the relationships between age at CI, audiological variables, and language outcomes. Finally, multivariate analysis was performed to quantify the relationship between It-Matrix, sentence recognition in quiet and at SNR+10 and TROG-2.

Results: The participants showed good speech recognition performance in quiet (94% for words and 89% for sentences) whilst their speech-in-noise scores decreased significantly. For the It-Matrix, only 9.2% of the participants showed scores within the normative range. This value was 60% for TROG-2 performance. For both auditory and language skills, group differences for unilateral versus bilateral CI users were not statistically significant ($p > 0.05$). Bivariate analysis showed that age at CI correlated significantly with overall results at TROG-2 ($r = -0.6$; $p < 0.001$) and with It-Matrix ($r = 0.5$; $p < 0.001$). TROG-2 was negatively correlated with results for It-Matrix ($r = -0.5$; $p < 0.001$). In the multivariate analysis with It-Matrix as a dependent variable, the model explained 63% of the variance, of which 60% was related to sentence recognition and 3% to morphosyntax.

Conclusions: These data contribute to the definition of average long-term outcomes expected in subjects implanted during childhood whilst increasing our knowledge of the effects of variables such as age at CI and morphosyntactic comprehension on speech perception. Although the majority of this prelingually DHH cohort did not achieve scores within a normative range, remarkably better It-Matrix scores were observed when compared to those from postlingually deafened adult CI users.

Keywords: cochlear implant, speech perception, deafness, adolescents, long-term outcomes, sentence recognition, listening condition

1. Introduction

Cochlear implants (CI) have been proven to be an effective technological source of treatment for children who have profound or severe hearing loss (deaf or hard of hearing, DHH) [1] and who get little or no benefit from hearing aids (HA). Indeed, several studies show that the majority of early implanted DHH children are able to achieve age-appropriate language skills during childhood [2–4]. On the other hand, many other factors appear to influence their postoperative outcomes, including age at implant [4–9] duration of auditory deprivation [4,10], bilateral listening [4,7], presence of multiple disabilities associated with hearing loss [4,7,11], caregivers' support [3,7,12], education and rehabilitative environment [4,13], intelligence quotient (IQ) [14,15], socio-economic factors [16], integrity of cochlear structures [4], surgical variables such as insertion depth and electrode type or CI signal processing [4].

As prelingually DHH pediatric CI users get older, questions regarding their long-term speech and language performance in adolescence or young adulthood arise and should be studied. In fact, most of the existing studies describe the short- or mid-term postoperative outcomes [2,5,9,17,18] obtained within the first 2-6 years of CI use, in particular regarding their auditory benefit and receptive/expressive language development. However, there are very few studies reporting on their long-term speech and language outcomes [19–21]. The first two papers by Uziel et al. [19] and Davidson et al. [20] reporting long-term outcomes in adolescents implanted in childhood were published more than ten years ago when speech perception tests based on adaptive paradigms were not as available as in recent years. Indeed, both studies used speech perception tests presented either in quiet or with a fixed signal-to-noise ratio (SNR).

So far, only one research article has been specifically designed to assess long-term outcomes in adolescents and young adults implanted during childhood, employing an adaptive noise test that is usually considered more relevant when assessing speech perception performance in settings which are similar to everyday complex listening environments [22]. Zalt et al. [22] studied speech-in-noise outcome differences between early and late implanted CI users, to examine the contribution of linguistic, cognitive, and background factors for speech perception in noise using the Hebrew Matrix sentence-in-noise test. Results showed poorer performance in noise when compared to hearing peers, with a large between-subject variability in the CI group. Matrix outcomes were found to be negatively correlated with non-verbal intelligence in a subgroup of adolescent CI users who showed greater than 50% correct word recognition in quiet.

The Italian version of the Matrix sentence test has been developed and validated in young hearing subjects for accurate and reliable speech recognition assessment in noise [23]. This test has been widely distributed and is currently used in research mainly for postlingually deaf adults [24,25].

However, no Matrix data for DHH adolescents and young adults implanted in childhood are available in the Italian language.

A further domain which is poorly understood is that of long-term morphosyntactic skills, an area of language acquisition which is considered an independent contributor to open-set speech perception [26,27]. It is fundamental when determining reading comprehension skills and remains at risk even in early implanted DHH children [28,29].

To our knowledge, no study has yet really assessed morphosyntactic competence in adolescents and young adults with a follow-up greater than 10 years. Moreover, there is a lack of studies in the Italian language exploring the relationship between speech perception in adaptive noise and language development in terms of morphosyntactic comprehension. Long-term assessments in prelingually DHH CI users are important inputs to help guide families and professionals in the therapeutic and counseling process and on the expectations and factors involved in the processes of developing the communication, educational and occupational skills of children who will grow up with CI.

Based on these premises, the primary aim of the present study was to assess the long-term speech perception outcomes for the adaptive Italian Matrix test (It-Matrix), and for word/sentence recognition in quiet and with fixed SNR, and morphosyntactic skills in a sample of unilaterally and bilaterally implanted adolescents and young adults, who received their CI during childhood. The secondary aim was to determine if and to what extent adaptive It-Matrix was correlated to speech perception in quiet and fixed SNR taking account of age at implantation and, furthermore, the extent to which language skills, in terms of morphosyntactic comprehension, were correlated in adulthood.

2. Materials and Methods

2.1 Study design and sample group

The present study was a retrospective cohort study involving prelingually DHH adolescents and young adults who received unilateral or bilateral CI during childhood assessed at the Cochlear Implant Centre of the xxxxxxxxxxxx. The data were analysed in accordance with the principles and later amendments to the Declaration of Helsinki (1964) and approved by the Hospital Ethics Committee (n. 259/2020).

Subjects were selected from a database of patients whose last follow-up occurred in the year prior to data collection. A total of 110 subjects were screened. Selection criteria for the current study included prelingual DHH subjects who had received CI during childhood, with a minimum period of 10 years of follow up after CI surgery, regular CI use (≥ 8 hours per day) and normo-typical development. The exclusion criteria were CI users with follow ups of less than 10 years, postlingually deaf CI users, partial-intermittent CI users, bimodal listeners, and the presence of associated neuropsychological disorders/disabilities.

Fifty-four prelingually DHH CI users (25 F, 29 M) satisfied the selection criteria and were included in the study. The average age at severe to profound sensorineural hearing loss diagnosis was 14.8 ± 9.8 months and the average age at CI surgery was 38.1 ± 24.6 months. The participants did not display any postoperative residual hearing in the implanted ear. Unilateral CI users showed a pure tone average (PTA) of 110.2 dB HL for octave frequencies below 1000 Hz in the non-implanted ear. None of the participants wore a contralateral HA. The average age at last follow-up assessment was 19.1 ± 4.3 years and the mean follow-up was 16 ± 3.7 years.

Deafness aetiology was unknown in 14 (26%) subjects, hereditary in 35 (65%) subjects (Connexin-26 gene mutation 79%, Usher syndrome 15%, Pendred Syndrome 3%, Waardenburg Syndrome 3%); and in 5 (9%) the cause of deafness was acquired (40% cytomegalovirus, 40% rubella virus; 20% meningitis). All but one of the subjects with Usher syndrome had mild night vision loss, and only one displayed daytime mild vison loss.

Thirty-three were unilateral, 21 were simultaneous (9) or sequential (12) bilateral CI users. Median sequential inter-implant time was 9 years (range 3 – 11).

Seven patients had a CI reimplantation: four patients from 90K, two patients from CI24RE and one from CI24M.

Thirty-three of the patients used Advanced Bionics (AB) devices (1 HiRes, 8 HiRes 120-S, 24 Optima-S) and twenty-one patients used Cochlear devices (21 ACE). All sound processors used in this study were Behind the Ear. Regarding unilateral users, 23/33 had all of the electrodes on, one had 5 electrodes off, one had 3 electrodes off, five had 2 electrodes off and three had 1 electrode off. Regarding bilateral users, 17/21 had all the electrodes on bilaterally, two had 2 electrodes off unilaterally and two had 2 electrodes off bilaterally.

All participants had used HAs prior to implantation and used spoken language as their primary mode of communication. All were native Italian speakers.

2.2 Retrospective data

Data were collected using our central clinical database where scores from tests performed during periodical follow up assessments were recorded. For each subject, the score recorded at the last assessment was used for the purposes of the study. Details of the reported data are described below for both audiological and language assessments.

2.3 Audiological assessment

Data from pure tone and speech perception testing in quiet and noise were used. Pure tone audiometry and speech perception testing was performed in a soundproof room using two loudspeakers (Indianaline, Coral electronic, Italy) positioned at 0° azimuth, 1 meter away from the patient's head when seated, connected to an audiometer (Aurical Aud, Otometrics, Natus Medical Srl, Italy) and using suitable laptop software (Otosuite, Otometrics, Natus Medical Srl, Italy).

PTA and sound field (SF) were both performed with warble tones for octave frequencies between 500 and 4000 Hz. Bilateral users were tested in daily listening mode with both ears.

Speech perception in quiet and fixed noise was evaluated in SF open set with two training lists of 20 bisyllabic words and two training lists of 10 sentences. The pre-recorded material of Cutugno et al. [30] was used and presented at 65 dB SPL in quiet and at fixed SNR +10 dB and +5 dB. The patients repeated words and sentences that they were able to understand, while the examiner evaluated the responses by assigning a score. For word lists, the performance score was based on a phonemic count, a word had 4 phonemic connections, with a minimum score set at 0 and the maximum at 4; for the lists of sentences two different scores were recorded: one score ranged from 0 to 3, since there were 3 keywords to repeat within the sentence; the second score was based on an evaluation of the full correct sentence. The program recorded the scores for each single performance and showed them as a percentage value.

Speech perception was also tested with adaptive noise through the Italian Matrix sentence test (It-Matrix) [23] which is the Italian adaptation of the Oldenburg Sentence Test (OLSA) [31]. The test consists of semantically unpredictable sentences but with a fixed syntactic structure: subject - verb - number - complement - adjective. The noise (speech noise) was presented at 65 dB SPL while the signal was adaptive. The examination was performed in all patients in open set: the patient repeated the words which they were able to understand, and the examiner marked the answers. The standardized and validated normal range data found from the second to the third adaptive measurement for Italian has a mean SRT of –6.7 ± 0.7 dB SNR and –7.4 ± 0.7 dB SNR for open- and closed-set tests. The results for It-Matrix were either a positive or negative dB value. The SRT represented the difference between the level of the speech signal and the level of the noise signal where the patient understood 50% of the words. As for the standardized procedure developed by Puglisi et al. [23], patients undertook two training lists of 30 items, followed by one test list of 30 items.

Both SF and speech perception testing in quiet and with fixed noise were completed by all subjects in the sample group, while 5 subjects were not able to complete the It-Matrix test. For these subjects, for statistical analysis purposes, the maximum test score (20 dB SNR), considered as the test limit, was computed [32].

2.4 Language assessment

As a measure of language competence, morphosyntactic comprehension was used. In our clinic, language competence is routinely assessed through the Italian standardized version of the Test for Reception of Grammar (TROG-2)[33]. TROG-2 consists of 20 blocks, each testing a specific grammatical construction, having an increasing order of difficulty. Each block contains four items and the child needs to respond correctly to all of them to level up. Each test stimulus is presented in a four-picture, multiple-choice format with lexical and grammatical foils. For each item, the examiner reads a sentence that refers to one of the four drawings, and the participant's task is to point to the drawing that corresponds to the meaning of the sentence. The raw score is calculated as a total number of achieved blocks and then converted into standard scores, using the tables included in the test manual. Based on its standard normative data, a score < 1 SD from the mean was considered as pathologic and this was indicated in the test manual as a standard score ≤ 85. Split-half reliability and internal consistency of the tests were 0.88 and 0.90, respectively.

The two tests were administered using spoken language to each CI subject individually in a quiet room, by one speech therapist.

2.5 Statistical Analysis

The Shapiro–Wilk test was used to assess normal data distribution. Categorical variables were calculated using frequencies and proportions whilst continuous data were estimated by means, standard deviations and ranges, where appropriate. The percentages of correct responses for speech perception in quiet and in noise were transformed to Rationalized Arcsine Units (RAUs) to limit the floor and ceiling effect [34].

A univariate analysis was performed using non-parametric tests. Kruskal-Wallis and Mann-Whitney U tests were used to compare listening modes (bilateral simultaneous, bilateral sequential, unilateral users), demographic variables (ages at diagnosis, HA, CI, last follow up and follow up length of time), audiological variables (SF, speech perception in quiet with fixed and adaptive noise) and finally linguistic variables (TROG-2). Where appropriate, average values for the study group were compared with those of the normative population with a one-sample z-test.

A bivariate analysis was conducted using the Spearman Rank Correlation Coefficient. It was used to calculate and investigate the relationships between age at CI, audiological variables (speech perception in quiet, with fixed and adaptive noise) and language (TROG-2) outcomes. A multivariate analysis was performed to quantify the relationships between a dependent variable (It-Matrix) and a set of explanatory variables (age at CI, speech perception in quiet and with noise, language skills, TROG-2) using a stepwise hierarchical linear regression model including all the variables with p ≤ 0.05 [35]. As noted below, the contribution of each variable to the prediction of the model was assessed in stages, progressively filtering the information, and allowing the identification of a statistically significant amount of variance in the outcomes that could be related to specific predictors. The variables that progressively entered the later stages of the analysis were tested for their specific contribution to variance after considering all the other preceding variables. A significant improvement in R^2 was achieved by comparing one model to another. Calculated p values were 2-sided, a *P*-value of less than 0.05 was considered as significant and the range of confidence interval was 95%, where appropriate. Statistical Analysis was performed using The Statistical Package for Social Sciences (SPSS) ver. 25 (SPSS IBM).

3. Results

3.1 Descriptive and comparative analysis

The mean SF at last follow up assessment was 31.3 ± 6.1 dB HL for the whole study group. Whole sample mean speech perception scores were as follows: words in quiet (W/Q) 94 ± 9.4%, words with SNR+10 (W/SNR+10) 71 ± 17.2%, words with SNR+5 dB (W/SNR+5) 42 ± 21%, sentences in quiet (S/Q) 89 ± 18%, sentences with SNR +10 dB (S/SNR+10) 64 ± 28% and sentences with SNR+5 dB (S/SNR+5) 26 ± 24%. Differences between speech in quiet and in noise were considered significant for both words and sentences (p<0.001).

Intelligibility in noise through It-Matrix showed a median SRT of -1.1 dB (range -6.8 – 20). This value is significantly different (p<0.001) to the normative mean of -6.8 (SD 0.8) dB SNR for the young, hearing population as reported by Puglisi et al [36]. Only 9.2% (5) of subjects fell within the normative range. Morphosyntactic comprehension assessed through TROG-2 showed a median score of 92 (range 55-119), with 60% of subjects scoring within the normal range of performance as reported in the manual (standard score ≥85). Table 1 reports detailed mean and median scores for audiological variables, speech perception (words and sentences in quiet, at fixed SNR +10 dB and +5 dB and It-Matrix) and morphosyntactic comprehension (TROG-2). Outcomes were separately reported in subgroups according to the subject's listening mode: bilateral (simultaneous/sequential) and unilateral.

Comparing each subgroup, the univariate comparative analysis showed statistically significant differences for all three listening modes concerning the following variables: age at last follow up (unilateral/bilateral simultaneous users p = 0.03; unilateral/bilateral sequential and bilateral simultaneous/bilateral sequential users p = <0.001); follow up length (p = <0.001; unilateral/bilateral sequential users p = <0.001; bilateral simultaneous/bilateral sequential users p = <0.001) and SF 500-

4000 Hz (unilateral/bilateral simultaneous users p = <0.001; unilateral/bilateral sequential users p = <0.001; bilateral simultaneous/bilateral sequential users p = <0.001). Considering bilateral users as just one sample, the only statistically significant difference with unilateral users was found for SF 500-4000 Hz (p = <0.001). No other significant differences were observed.

3.2 Correlations between age at CI, audiological variables and morphosyntax

Table 2 shows results from bivariate analysis. Overall age at CI was strongly correlated with age at last follow-up, with sentence recognition in quiet and at SNR +10, It-Matrix, and TROG-2. It-Matrix was strongly correlated with speech perception in quiet and fixed SNR (p <0.001). Considering the large datasets of outcome variables and the high correlations between them, to reduce the number of features per observation a Principal Component Analysis (PCA) was used [37]. Two PCAs were identified regarding speech perception: the PCA word recognition (PCA-W) and the PCA sentence recognition (PCA-S), both based on the RAU scores. Both PCAs were based on open-set recognition scores in quiet and in fixed noise presented at SNR +10 dB and SNR +5 dB (Table 3). Both PCAs had good loading of components; KMO index was equal to 0.64 for PCA-W and to 0.60 for PCA-S, while for both, the Bartlett's test was statistically significant with $p < 0.001$.

The new bivariate (Table 4) showed that It-Matrix was significantly correlated with all speech perception outcomes and with morphosyntax (TROG-2), and the significant correlations continued while controlling for age at CI and for age at last follow up. Furthermore, both It-Matrix and TROG-2 maintained their significant correlations with age at CI while controlling for age at last follow-up. All correlations showed a medium/good coefficient.

3.3 Factors predicting It-Matrix

The multivariate regression analysis was performed stepwise including variables from bivariate partial correlation and comparative analysis with $p \leq 0.05$.

Table 5 shows the analysis using It-Matrix as the dependent variable. Step 1 included Age at CI and Age at Last Follow Up and the model was entirely explained by Age at CI alone that accounted for almost 40% of the variance. In step 2, PCA-W and PCA-S were computed to the model, adding a further 21% to the overall variance, thereby reaching 61% of explained variance. In this second step the model variance was explained by PCA-S, while age at CI was excluded by the model. In step 3, TROG-2, which described morphosyntactic comprehension, was added and apportioned an additional 3.6%, for a total 65% of explainable variances. This model suggests that there is a specific contribution of PCA-S and TROG-2 to It-Matrix.

4. Discussion

Long-term assessments of implanted children are important inputs to guide families and professionals in therapeutic and counselling processes. There are relatively few studies reporting on long-term outcomes in adolescents and young adults implanted during childhood, and who have grown up with CI. Hence the present study aimed to assess the long-term speech and language outcomes in a sample of prelingually DHH adolescents and young adults.

4.1 Primary outcome: Long-term speech perception outcomes and morphosyntactic comprehension

Speech perception tests in quiet are being used routinely for the assessment of performance benefits in pediatric and adult CI users. CI users generally demonstrate higher level performance in quiet listening conditions. Although these test materials provide valuable information regarding users' performance, they do not give realistic information for adverse conditions such as listening in the presence of background noise at different SNRs [38]. The present study provided, for the first time, the long-term outcomes for speech perception in quiet and in noise tests conducted in Italian, measured in a heterogeneous sample of adolescents and young adults implanted during childhood. One primary outcome of the present study was to evaluate long-term speech perception of words and sentences in quiet and with fixed SNR, and in adaptive noise using the Italian adaptation of the Oldenburg sentence test (It-Matrix). The data that emerged in our study showed that the use of CI positively contributed to the recognition of sentences in quiet and in noise, even in children with late access to surgery. However, the average values for speech perception in adaptive noise were significantly worse when compared to normative values, and only 9.2% of subjects fell within the normative range. Accordingly, words and sentence recognition at SNR + 10 dB and +5 dB showed a noteworthy performance deterioration compared to speech tests in quiet.

There is a paucity of papers reporting outcomes for fixed SNR and adaptive noise tests collected over the longer term. As mentioned above, the Uziel et al. study [19] involving 79 adolescents reported mean word recognition in quiet and with SNR+10 dB of 72% and 44% respectively. Age at CI was found to be a strong predictor of outcomes. Davidson et al. [20] performed a study observing mid- and long-term outcomes in a group of 112 teenagers. The authors observed that open-set recognition scores for words and sentences increased significantly as a function of increased age and listening experience: word recognition in quiet was 60% and sentences recognition in quiet and in noise were 80% and 52%, respectively. Outcomes recorded in the present study were only slightly better being 94% and 71% for words in quiet and with SNR +10 dB, and 89% and 64% for sentences in quiet and with SNR +10 dB. These differences might be explained by the different test materials, but also by a higher mean age at test and a longer follow-up for subjects belonging to the present study. As discussed by Davidson et al. [20] and Beadle et al. [39] speech perception and language skills tend to improve with age, positively influenced by increased experience with CI and by taking advantage of improved linguistic competence [40].

Regarding the current literature on It-Matrix sentence tests, there are currently two articles on adults and only one article on pediatric subjects. Gallo et al. [24] explored It-Matrix in a cohort of 45 unilateral and bimodal CI users, with a median age at test of 50 years and with short-term CI use. In their sample, unilateral and bimodal users achieved a median SRT of 4.15 dB and 2.85 dB respectively, with a wide range of outcomes. Dincer D'Alessandro et al.[25] assessed a group of 20 unilateral CI users with a median age of 65, and a median SRT of 7.6 dB SNR, once again with wide intrasubject variability. Concerning the pediatric population, Forli et al. [41] analyzed a sample of 36 children with bilateral sequential CI and a mean age of 11 years and a mean inter-implant time of 5 years. The mean SRTs reported were 3.9 dB with the first CI and 2 dB with the bilateral sequential CI respectively, which were found to be significantly different. Overall, the results from the above studies show large variability dependent on the sample examined (age, audiological characteristics, listening mode) and on the assessment setting.

It-Matrix intelligibility in our sample population showed a median SRT of -1.1 dB SNR for the whole sample and a value of -0.6 dB, -2 dB and -1.7 dB SNR for unilateral, bilateral sequential and bilateral simultaneous users respectively. These results are considerably better when compared to the studies of the adult population by Gallo et al. and Dincer D'Alessandro et al. [24,25] but also when compared to the study on younger subjects by Forli et al. [41].

Concerning better outcomes for It-Matrix observed in our study when compared to the adult hearing population, different variables might support such differences. From a perceptive point of view, speech perception through CI is influenced by frequency resolution which is poorer in CI when compared to hearing subjects [42]. Nevertheless, poor frequency resolution does not appear to limit speech perception in noise for prelingually deaf, early implanted children as much as it does for postlingually deaf adults. Short-term hearing deprivation and brain plasticity [43], which was observed in most children included in the present study, might have supported better outcomes as opposed to the adult population from Gallo et al.[24].

When comparing It-Matrix results to those found in the early implanted pediatric population by Forli et al.[41], better outcomes were observed for subjects included in the present study. Performance differences with worse outcomes reported by Forli et al. [41] might be due to younger mean age at test and the wider age range of subjects included in their work, whereas the standard It-Matrix test was administered in a younger pediatric sample (≥ 6 years). This once again is possibly related to the influence that age has on speech perception score, which increases significantly as a function of increased age and listening experience, being positively influenced by increased CI experience [20,39].

Children with different listening modes showed significantly better results for bilateral listeners in SF audiometric threshold, possibly owing to the summation effect measured in the current S0/N0 setting. Better SF thresholds increase audibility that in turn has been shown to be clinically more robust in bilateral when compared to unilateral and bimodal pediatric CI users, and in turn positively influence speech perception outcomes [44].

Sequential and simultaneous bilateral users did not show significant differences for speech perception in quiet and in noise. Most likely, such results reflect the fact that our sample included a high percentage of subjects with sequential implants (> 4 years) and there were no substantial age-related group differences at CI age. However, it may be argued that in the long term, as suggested by Kim et al. [45] the effects of the delayed sequential implantation might not be as relevant when speech discrimination is assessed in the S0/N0 testing mode. This last outcome once again might be linked to the prevalence of unilateral users in the study group and to the prevalence of subjects with sequential implantation in the bilateral subgroup. The composition of the sample size was determined by a more recent diffusion of the UNHS in Italy [46] and by differences in funding models between countries [47]. Therefore, there are still many patients who received unilateral implant or delayed sequential bilateral implant in childhood who are now young adults. Finally, although group differences between unilateral and bilateral users (1.4 dB SNR) were not statistically significant at It-Matrix, differences higher than 1 dB SNR are considered clinically significant [48]

Morphosyntactic comprehension is considered an independent contributor to open-set speech perception in paediatric HA or CI users [26,27]. Furthermore, morphosyntactic knowledge is fundamental when determining reading comprehension skills in DHH subjects and could be considered one of the most important factors that explain the variances observed in DHH students [29]. The present sample showed wide inter-individual variability for this skill. Although the percentage of subjects within the normal range (a score ≥ 85) was 60%, the other 40% didn't achieve normal syntactic comprehension despite a mean period of 16 years of CI use. Indeed, children with higher scores in this kind of task have probably developed the skill set necessary to recognize entire sentences with the support of syntactic knowledge [28].

During childhood, several studies highlighted the risk for CI users to show fragility in the domain of morphosyntactic skills, both in the expressive and comprehension areas [49–55]. CI recipient children tend to reveal poorer grammatical processing such as omissions or substitutions of clitic pronouns marking gender or number, and verbal flexions [50]. They also show poorer performance when using complex sentence structures and such outcome differences with hearing peers are still present after 5 years of CI use [51].

As for morphosyntactic production, variability in results was also reported in comprehension [52–55]. Regarding this aspect, Geers et al. [54] found that about two thirds of CI recipient children aged 5–7 years and implanted within 5 years of chronological age, scored one standard deviation or more below controls on language comprehension. Schorr et al. [53] reported similar results for CI recipient children aged 5–14 years (age at implant 0;11 to 5;1), although this proportion decreases to 40% in Geers et al. [52], and down to one third in Hansson et al. [55].

The present study is, at this time, the only one that has been published concerning morphosyntactic comprehension of CI adolescents and young adults who were implanted during childhood. Therefore, it is not possible to make a direct comparison with other studies. If we compare our findings with the above reported literature, it suggests that the proportion of CI subjects with problems in morphosyntactic comprehension to be about 30%-40% as already indicated by Geers et al. and Hansson et al. [52,55], and this deficit can still be observed after 16 or more years of CI use and during adulthood. These results differ from those which were reported by Breland et al. [56], who found that differences detectable during earlier years of school may disappear during the high-school period. The present study included subjects who were implanted more than 20 years ago, when diagnosis of hearing loss and intervention was more delayed when compared to more recent years. Early intervention is one of the primary acknowledged factors impacting linguistic outcomes [57] and the present findings showed significant effects of age at implantation on morphosyntactic development.

Finally, statistically significant differences were not observed in our study even when dividing subjects into subgroups according to listening mode. Also, in this case, as discussed in the speech perception section, it could possibly be due to the smaller sample size and higher number of sequential versus simultaneous patients. In fact, other studies have shown better outcomes in bilateral young adult users with at least 10 years use regarding the development of receptive and expressive outcomes when compared to their peers with unilateral CI [58–60].

4.2 Secondary outcome: correlations between subjective, audiological variables and morphosyntactic comprehension

The bivariate analysis showed a significant correlation between speech perception and age at implantation especially in difficult listening conditions: the effects of age at CI persist into adulthood and both speech perception at fixed SNR and It-Matrix were inversely correlated with age at CI even when data were adjusted for age at test.

These results are in line with those described by Zalt et al. [22] in their long-term case control study. The authors found significant differences in outcomes for speech perception in quiet and for the Hebrew Matrix in all subgroups when compared to hearing controls. These results were mainly influenced by age at diagnosis and implantation despite a large subjective variability. Also, Pearson analysis showed a significant correlation between speech-in-noise and Raven, the receptive vocabulary, and the phonemic fluency score, underlying once again how speech perception is highly correlated to language.

Furthermore, our study highlights the correlation between the It-Matrix results and recognition of words and sentences in quiet and in fixed signal noise and this outcome is not influenced by age at CI or age at test. In general, It-Matrix results for the whole sample had great interpersonal and intergroup variability with similar results being found by Zaltz et al.[22] (SRT values between -4.5 and +1.25; a range of approximately 17 dB SNR). In addition, we demonstrated no differences between unilateral or bilateral samples.

The correlation of It-Matrix with speech perception in noise with fixed SNR +5 dB and +10 dB is a further novelty of the present study, as there is no other study which confirms this correlation both in the Italian language and for young CI users. Only a few studies have compared two or more different sentence-level SIN tests in the hearing-impaired adult population [61–63]. In particular, Jansen et al. [62] observed how the French Matrix had a significant correlation with everyday sentences in noise tests showing that the newly developed test was accurate and reliable in a large group of hearing and DHH subjects. In our opinion, the presence of a strong correlation between the standard It-Matrix and the Italian standard words and sentences in noise tests supports reliability and accuracy of measurements in the adolescent and young adult early implanted population.

Concerning morphosyntactic comprehension, this skill was found to be strongly correlated to It-Matrix and age at implantation. However, using a stepwise multiple regression analysis the weight of age at the time of implant decreased when predicting performance over It-Matrix and morphosyntactic comprehension. Late implantation age is therefore a risk factor, but it does not of itself explain the differences in auditory perception in noise or language development. Regarding the It-Matrix test, the most significant variables were PCA-S and TROG-2 (morphosyntactic comprehension). PCA-S alone explained 60% of variance and TROG-2 added 3% to the total variance. The ability to adequately perceive and repeat sentences, both in quiet and noise, was therefore the greater contributor to CI users' performance in listening with adaptive noise, probably due to the similarity between all of these tasks where the subject is asked to repeat a series of words, maintaining not only their semantic meaning, as happens for word recognition tasks, but also processing their role and function in sentence structure. This is an ability that, with respect to word recognition tasks, also requires more mature and efficient competences regarding other cognitive factors, such as working memory. In fact, in a study on postlingually deafened adult CI users, Kaandorp et al. [64] observed that the working memory capacity alone, measured through the Reading Span, explained 55% of the variance in SIN thresholds, suggesting that poor verbal working memory capacity limits speech-recognition abilities in CI listeners. In the same way, working memory may also influence performance in adults who grew up with CI and this facet should receive more attention.

Regarding the role of linguistic aspects, which, in the present study, were represented by the scores obtained at TROG-2. Various authors have already probed the independent contributions of speech production and language ability to open-set speech perception scores in children using either HAs or CIs [26,27]. Furthermore, Eisenberg et al. [28] found that children's performance in Comprehensive Assessment of Spoken Language, a test structured to assess subjects' skills in syntactic constructions and paragraph comprehension, yielded the strongest correlation with sentence recognition. They concluded that children with higher scores in this type of syntactic task have probably developed, at the same time, the skill set necessary to recognize entire sentences with the support of syntactic knowledge and this phenomenon is already detectable commencing 5-6 years after CI activation. Our study supports the hypothesis that syntactic skills are still influential in adulthood, but with a smaller contribution than that measured by Eisenberg et al. in the pediatric population [28].

Limitations and future directions

A limitation of the present study concerns the paucity of bilateral simultaneous users in the study sample. Because of the small sample size and the absence of differences in age range at implantation between sequential and concurrent users, a comparative analysis was performed but was not statistically significant. However, it is possible that by supplementing the sample of simultaneous bilateral users with the results of the next generation of implanted children, these results may change, as nowadays simultaneous bilateral implantation is becoming an earlier and more widespread indication than in the past. Another limitation of the present study is the fact that, due to the retrospective nature of data collection, we were unable to test for other variables that could have explained the 40% residual variance. One of these variables is non-verbal intelligence, which has been found to be significantly correlated with Matrix outcomes in adolescent and young adult CI users by Zaltz et al. [22]. Conversely, non-verbal intelligence was not found to correlate significantly with adaptive speech perception in noise outcomes in CI recipient children with typical and atypical language development by Torkildsen et al. [65].

Non-verbal intelligence has been found to correlate with receptive and expressive language skills in children with associated disabilities [14]. In contrast to the Meinzen-Derr study, non-verbal intelligence didn't reach statistical significance within a model of linear correlation including early CI and socioeconomic status. Particularly early implantation remains a dominant factor in children and adolescents, leading to better outcomes [66]. Similarly, the effects of non-verbal intelligence in a large cohort of early implanted children showed no significant correlations with postoperative comprehensive assessments of spoken language [67].

Differing results concerning the effects of non-verbal IQ between various studies might be as a result of different test methodologies and the lack of normative data for children with hearing loss, disadvantaging them in terms of clinical interpretation and recommendations [67].

The present study, being retrospective in nature, did not have complete non-verbal intelligence data, and therefore, could not contribute to shedding light on this important aspect.

Nevertheless, the outcomes of this study have provided a solid foundation for future prospective research, which should be designed to verify the role of variables such as auditory attention, working memory and cognitive functions on speech perception in noise and linguistic skills.

Conclusions

The results of the present study show the positive contribution made by CI in speech perception in quiet and in noise and in morphosyntactic skills, in DHH adolescents and young adults who received CI in childhood. Performances in quiet and in noise with fixed and adaptive SNR were, on average, better than that reported in the DHH adult population, despite wide variability in individual performances. Age at CI still represents an important factor influencing outcomes in long-term assessments. The study also provides insight into how speech perception and morphosyntactic comprehension are still interlinked in adolescence and adulthood.

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Long-term speech perception and morphosyntactic outcomes in adolescents and young adults implanted in childhood

Abstract

Background: Long-term assessments of ~~speech perception and language outcomes in children~~ children with cochlear implants (CI) are important inputs to help guide families and professionals in therapeutic and counselling processes. Based on these premises, the primary aim of the present study was to assess the long-term speech and language outcomes in a sample of prelingually deaf or hard of hearing (DHH) adolescents and young adults with unilateral or bilateral implantation in childhood. The secondary aim was to investigate the correlations of age at implantation with long-term speech and language outcomes.

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Materials and Methods: Retrospective observational study on 54 long-term CI users, 33 unilateral and 21 bilateral (mean age at CI surgery 38.1 ± 24.6 months; mean age at last follow-up assessment 19.1 ± 4.3 years of age and mean follow-up time 16 ± 3.7 years). Means and standards were used to describe speech perception (in quiet, in fixed noise and in adaptive noise using It-Matrix) and morphosyntactic comprehension (TROG-2) outcomes. A univariate analysis was used to evaluate outcome differences between unilateral and bilateral patients. Bivariate analysis was performed to investigate the relationships between age at CI, audiological variables, and language outcomes. Finally, multivariate analysis was performed to quantify the relationship between It-Matrix, sentence recognition in quiet and at SNR+10 and TROG-2.

Results: **The participants showed good speech recognition performance in quiet (94% for words and 89% for sentences) whilst their speech-in-noise scores decreased significantly. For the It-Matrix, only 9.2% of the participants showed scores within the normative range. This value was 60% for TROG-2 performance. For both auditory and language skills, group differences for unilateral versus bilateral CI users were not statistically significant ($p > 0.05$).** Bivariate analysis showed that age at CI correlated significantly with overall results at TROG-2 ($r = -0.6$; $p < 0.001$) and with It-Matrix ($r = 0.5$; $p < 0.001$). TROG-2 was negatively correlated with results for It-Matrix ($r = -0.5$; $p < 0.001$). In the multivariate analysis with It-Matrix as a dependent variable, the model explained 63% of the variance, of which 60% was related to sentence recognition and 3% to morphosyntax.

Conclusions: These data contribute to the definition of average long-term outcomes expected in subjects implanted during childhood whilst increasing our knowledge of the effects of variables such as age at CI and morphosyntactic comprehension on speech perception. **Although the majority of this prelingually DHH cohort did not achieve scores within a normative range, remarkably**

better It-Matrix scores were observed when compared to those from postlingually deafened adult CI users.

Keywords: cochlear implant, speech perception, deafness, adolescents, long-term outcomes, sentence recognition, listening condition

1. Introduction

Cochlear implants (CI) have been proven to be an effective technological source of treatment for **children who have profound or severe hearing loss (deaf or hard of hearing, DHH))** [1]**and who get little or no benefit from hearing aids (HA).** Indeed, several studies show that the majority of early implanted DHH children are able to achieve age-appropriate language skills during childhood [2–4][1–3]. On the other hand, many other factors appear to influence their postoperative outcomes, including age at implant [4–9][3–8] duration of auditory deprivation [4,10][3,9], bilateral listening [4,7][3,6], presence of multiple disabilities associated with hearing loss [4,7,11][3,6,10], caregivers' support [3,7,12][2,6,11], education and rehabilitative environment [4,13][3,12], intelligence quotient (IQ) [14,15][13,14], socio-economic factors [16][15], integrity of cochlear structures [4][3], surgical variables such as insertion depth and electrode type or CI signal processing [4][3].

As prelingually DHH pediatric CI users get older, questions regarding their long-term speech and language performance in adolescence or young adulthood arise and should be studied. In fact, most of the existing studies describe the short- or mid-term postoperative outcomes [2,5,9,17,18][1,4,8,16,17] obtained within the first 2-6 years of CI use, in particular regarding their auditory benefit and receptive/expressive language development. However, there are very few studies reporting on their long-term speech and language outcomes [19–21][18–20]. The first two papers by Uziel et al. [19][18] and Davidson et al. [20][19] reporting long-term outcomes in adolescents implanted in childhood were published more than ten years ago when speech perception tests based on adaptive paradigms were not as available as in recent years. Indeed, both studies used speech perception tests presented either in quiet or with a fixed signal-to-noise ratio (SNR).

So far, only one research article has been specifically designed to assess long-term outcomes in adolescents and young adults implanted during childhood, employing an adaptive noise test that is usually considered more relevant when assessing speech perception performance in settings which are similar to everyday complex listening environments [22][21]. Zalt et al. [22][21] studied speech-in-noise outcome differences between early and late implanted CI users, to examine the contribution of linguistic, cognitive, and background factors for speech perception in noise using the Hebrew Matrix sentence-in-noise test. Results showed poorer performance in noise when compared to hearing peers, with a large between-subject variability in the CI group. Matrix outcomes were found to be negatively correlated with non-verbal intelligence in a subgroup of adolescent CI users who showed greater than 50% correct word recognition in quiet.

The Italian version of the Matrix sentence test has been developed and validated in young hearing subjects for accurate and reliable speech recognition assessment in noise [23][22]. This test has been widely distributed and is currently used in research mainly for postlingually deaf adults

[24,25]~~[23,24]~~. However, no Matrix data for DHH adolescents and young adults implanted in childhood are available in the Italian language.

A further domain which is poorly understood is that of long-term morphosyntactic skills, an area of language acquisition which is considered an independent contributor to open-set speech perception [26,27]~~**[26,27]**~~**. It is fundamental when determining reading comprehension skills and remains at risk even in early implanted DHH children** [28,29]**[28,29].**

To our knowledge, no study has yet really assessed morphosyntactic competence in adolescents and young adults with a follow-up greater than 10 years. Moreover, there is a lack of studies in the Italian language exploring the relationship between speech perception in adaptive noise and language development in terms of morphosyntactic comprehension. Long-term assessments in prelingually DHH CI users are important inputs to help guide families and professionals in the therapeutic and counseling process and on the expectations and factors involved in the processes of developing the communication, educational and occupational skills of children who will grow up with CI.

Based on these premises, the primary aim of the present study was to assess the long-term speech perception outcomes for the adaptive Italian Matrix test (It-Matrix), and for word/sentence recognition in quiet and with fixed SNR, and morphosyntactic skills in a sample of unilaterally and bilaterally implanted adolescents and young adults, who received their CI during childhood. The secondary aim was to determine if and to what extent adaptive It-Matrix was correlated to speech perception in quiet and fixed SNR taking account of age at implantation and, furthermore, the extent to which language skills, in terms of morphosyntactic comprehension, were correlated in adulthood.

2. Materials and Methods

2.1 Study design and sample group

The present study was a retrospective cohort study involving prelingually DHH adolescents and young adults who received unilateral or bilateral CI during childhood assessed at the Cochlear Implant Centre of the xxxxxxxxxxxx. The data were analysed in accordance with the principles and later amendments to the Declaration of Helsinki (1964) and approved by the Hospital Ethics Committee (n. 259/2020).

Subjects were selected from a database of patients whose last follow-up occurred in the year prior to data collection. A total of 110 subjects were screened. Selection criteria for the current study included prelingual DHH subjects who had received CI during childhood, with a minimum period of 10 years of follow up after CI surgery, regular CI use (≥ 8 hours per day) and normo-typical development. The exclusion criteria were CI users with follow ups of less than 10 years, postlingually deaf CI users, partial-intermittent CI users, bimodal listeners, and the presence of associated neuropsychological disorders/disabilities.

Fifty-four prelingually DHH CI users (25 F, 29 M) satisfied the selection criteria and were included in the study. The average age at severe to profound sensorineural hearing loss diagnosis was 14.8 ± 9.8 months and the average age at CI surgery was 38.1 ± 24.6 months. **The participants did not display any postoperative residual hearing in the implanted ear. Unilateral CI users showed a pure tone average (PTA) of 110.2 dB HL for octave frequencies below 1000 Hz in the non-implanted ear. None of the participants wore a contralateral HA.** The average age at last follow-up assessment was 19.1 ± 4.3 years and the mean follow-up was 16 ± 3.7 years.

Deafness aetiology was unknown in 14 (26%) subjects, hereditary in 35 (65%) subjects (Connexin-26 gene mutation 79%, Usher syndrome 15%, Pendred Syndrome 3%, Waardenburg Syndrome 3%); and in 5 (9%) the cause of deafness was acquired (40% cytomegalovirus, 40% rubella virus; 20% meningitis). All but one of the subjects with Usher syndrome had mild night vision loss, and only one displayed daytime mild vison loss.

Thirty-three were unilateral, 21 were simultaneous (9) or sequential (12) bilateral CI users. Median sequential inter-implant time was 9 years (range 3 – 11).

Seven patients had a CI reimplantation: four patients from 90K, two patients from CI24RE and one from CI24M.

Thirty-three of the patients used Advanced Bionics (AB) devices (1 HiRes, 8 HiRes 120-S, 24 Optima-S) and twenty-one patients used Cochlear devices (21 ACE). All sound processors used in this study were Behind the Ear. Regarding unilateral users, 23/33 had all of the electrodes on, one had 5 electrodes off, one had 3 electrodes off, five had 2 electrodes off and three had 1 electrode off. Regarding bilateral users, 17/21 had all the electrodes on bilaterally, two had 2 electrodes off unilaterally and two had 2 electrodes off bilaterally.

All participants had used HAs prior to implantation and used spoken language as their primary mode of communication. All were native Italian speakers.

2.2 Retrospective data

Data were collected using our central clinical database where scores from tests performed during periodical follow up assessments were recorded. For each subject, the score recorded at the last assessment was used for the purposes of the study. Details of the reported data are described below for both audiological and language assessments.

2.3 Audiological assessment

Data from pure tone and speech perception testing in quiet and noise were used. Pure tone audiometry and speech perception testing was performed in a soundproof room using two loudspeakers (Indianaline, Coral electronic, Italy) positioned at 0° azimuth, 1 meter away from the patient's head when seated, connected to an audiometer (Aurical Aud, Otometrics, Natus Medical Srl, Italy) and using suitable laptop software (Otosuite, Otometrics, Natus Medical Srl, Italy).

PTA and sound field (SF) were both performed with warble tones for octave frequencies between 500 and 4000 Hz. Bilateral users were tested in daily listening mode with both ears.

Speech perception in quiet and fixed noise was evaluated in SF open set with two training lists of 20 bisyllabic words and two training lists of 10 sentences. The pre-recorded material of Cutugno et al. [30][30] was used and presented at 65 dB SPL in quiet and at fixed SNR +10 dB and +5 dB. The patients repeated words and sentences that they were able to understand, while the examiner evaluated the responses by assigning a score. For word lists, the performance score was based on a phonemic count, a word had 4 phonemic connections, with a minimum score set at 0 and the maximum at 4; for the lists of sentences two different scores were recorded: one score ranged from 0 to 3, since there were 3 keywords to repeat within the sentence; the second score was based on an evaluation of the full correct sentence. The program recorded the scores for each single performance and showed them as a percentage value.

Speech perception was also tested with adaptive noise through the Italian Matrix sentence test (It-Matrix) [23][22] which is the Italian adaptation of the Oldenburg Sentence Test (OLSA) [31][31]. The test consists of semantically unpredictable sentences but with a fixed syntactic structure: subject - verb - number - complement - adjective. The noise (speech noise) was presented at 65 dB SPL while the signal was adaptive. The examination was performed in all patients in open set: the patient repeated the words which they were able to understand, and the examiner marked the answers. The standardized and validated normal range data found from the second to the third adaptive measurement for Italian has a mean SRT of –6.7 ± 0.7 dB SNR and –7.4 ± 0.7 dB SNR for open- and closed-set tests. The results for It-Matrix were either a positive or negative dB value. The SRT represented the difference between the level of the speech signal and the level of the noise signal where the patient understood 50% of the words. As for the standardized procedure developed by Puglisi et al. [23][22], patients undertook two training lists of 30 items, followed by one test list of 30 items.

Both SF and speech perception testing in quiet and with fixed noise were completed by all subjects in the sample group, while 5 subjects were not able to complete the It-Matrix test. For these subjects, for statistical analysis purposes, the maximum test score (20 dB SNR), considered as the test limit, was computed [32][32].

2.4 Language assessment

As a measure of language competence, morphosyntactic comprehension was used. In our clinic, language competence is routinely assessed through the Italian standardized version of the Test for Reception of Grammar (TROG-2)[33][33]. TROG-2 consists of 20 blocks, each testing a specific grammatical construction, having an increasing order of difficulty. Each block contains four items and the child needs to respond correctly to all of them to level up. Each test stimulus is presented in a four-picture, multiple-choice format with lexical and grammatical foils. For each item, the examiner reads a sentence that refers to one of the four drawings, and the participant's task is to point to the drawing that corresponds to the meaning of the sentence. The raw score is calculated as a total number of achieved blocks and then converted into standard scores, using the tables included in the test manual. Based on its standard normative data, a score < 1 SD from the mean was considered as pathologic and this was indicated in the test manual as a standard score ≤ 85. Split-half reliability and internal consistency of the tests were 0.88 and 0.90, respectively.

The two tests were administered using spoken language to each CI subject individually in a quiet room, by one speech therapist.

2.5 Statistical Analysis

The Shapiro–Wilk test was used to assess normal data distribution. Categorical variables were calculated using frequencies and proportions whilst continuous data were estimated by means, standard deviations and ranges, where appropriate. The percentages of correct responses for speech perception in quiet and in noise were transformed to Rationalized Arcsine Units (RAUs) to limit the floor and ceiling effect [34][34].

A univariate analysis was performed using non-parametric tests. Kruskal-Wallis and Mann-Whitney U tests were used to compare listening modes (bilateral simultaneous, bilateral sequential, unilateral users), demographic variables (ages at diagnosis, HA, CI, last follow up and follow up length of time), audiological variables (SF, speech perception in quiet with fixed and adaptive noise) and finally linguistic variables (TROG-2). Where appropriate, average values for the study group were compared with those of the normative population with a one-sample z-test.

A bivariate analysis was conducted using the Spearman Rank Correlation Coefficient. It was used to calculate and investigate the relationships between age at CI, audiological variables (speech perception in quiet, with fixed and adaptive noise) and language (TROG-2) outcomes. A multivariate analysis was performed to quantify the relationships between a dependent variable (It-Matrix) and a set of explanatory variables (age at CI, speech perception in quiet and with noise, language skills, TROG-2) using a stepwise hierarchical linear regression model including all the variables with p ≤ 0.05 [35][35]. As noted below, the contribution of each variable to the prediction of the model was assessed in stages, progressively filtering the information, and allowing the identification of a statistically significant amount of variance in the outcomes that could be related to specific predictors. The variables that progressively entered the later stages of the analysis were tested for their specific contribution to variance after considering all the other preceding variables. A significant improvement in R^2 was achieved by comparing one model to another. Calculated p values were 2-sided, a *P*-value of less than 0.05 was considered as significant and the range of confidence interval was 95%, where appropriate. Statistical Analysis was performed using The Statistical Package for Social Sciences (SPSS) ver. 25 (SPSS IBM).

3. Results

3.1 Descriptive and comparative analysis

The mean SF at last follow up assessment was 31.3 ± 6.1 dB HL for the whole study group. Whole sample mean speech perception scores were as follows: words in quiet (W/Q) 94 ± 9.4%, words with SNR+10 (W/SNR+10) 71 ± 17.2%, words with SNR+5 dB (W/SNR+5) 42 ± 21%, sentences in quiet (S/Q) 89 ± 18%, sentences with SNR +10 dB (S/SNR+10) 64 ± 28% and sentences with SNR+5 dB (S/SNR+5) 26 ± 24%. Differences between speech in quiet and in noise were considered significant for both words and sentences (p<0.001).

Intelligibility in noise through It-Matrix showed a median SRT of -1.1 dB (range -6.8 – 20). This value is significantly different (p<0.001) to the normative mean of -6.8 (SD 0.8) dB SNR for the young, hearing population as reported by Puglisi et al [36][36]. Only 9.2% (5) of subjects fell within the normative range. Morphosyntactic comprehension assessed through TROG-2 showed a median score of 92 (range 55-119), with 60% of subjects scoring within the normal range of performance as reported in the manual (standard score ≥85). Table 1 reports detailed mean and median scores for audiological variables, speech perception (words and sentences in quiet, at fixed SNR +10 dB and +5 dB and It-Matrix) and morphosyntactic comprehension (TROG-2). Outcomes were separately reported in subgroups according to the subject’s listening mode: bilateral (simultaneous/sequential) and unilateral.

Comparing each subgroup, the univariate comparative analysis showed statistically significant differences for all three listening modes concerning the following variables: age at last follow up (unilateral/bilateral simultaneous users p = 0.03; unilateral/bilateral sequential and bilateral simultaneous/bilateral sequential users p = <0.001); follow up length (p = <0.001; unilateral/bilateral sequential users p = <0.001; bilateral simultaneous/bilateral sequential users p = <0.001) and SF 500-

4000 Hz (unilateral/bilateral simultaneous users p = <0.001; unilateral/bilateral sequential users p = <0.001; bilateral simultaneous/bilateral sequential users p = <0.001). Considering bilateral users as just one sample, the only statistically significant difference with unilateral users was found for SF 500-4000 Hz (p = <0.001). No other significant differences were observed.

3.2 Correlations between age at CI, audiological variables and morphosyntax

Table 2 shows results from bivariate analysis. Overall age at CI was strongly correlated with age at last follow-up, with sentence recognition in quiet and at SNR +10, It-Matrix, and TROG-2. It-Matrix was strongly correlated with speech perception in quiet and fixed SNR (p <0.001). Considering the large datasets of outcome variables and the high correlations between them, to reduce the number of features per observation a Principal Component Analysis (PCA) was used [37]~~[37]~~. Two PCAs were identified regarding speech perception: the PCA word recognition (PCA-W) and the PCA sentence recognition (PCA-S), both based on the RAU scores. Both PCAs were based on open-set recognition scores in quiet and in fixed noise presented at SNR +10 dB and SNR +5 dB (Table 3). Both PCAs had good loading of components; KMO index was equal to 0.64 for PCA-W and to 0.60 for PCA-S, while for both, the Bartlett's test was statistically significant with p < 0.001.

The new bivariate (Table 4) showed that It-Matrix was significantly correlated with all speech perception outcomes and with morphosyntax (TROG-2), and the significant correlations continued while controlling for age at CI and for age at last follow up. Furthermore, both It-Matrix and TROG-2 maintained their significant correlations with age at CI while controlling for age at last follow-up. All correlations showed a medium/good coefficient.

3.3 Factors predicting It-Matrix

The multivariate regression analysis was performed stepwise including variables from bivariate partial correlation and comparative analysis with p ≤ 0.05.

Table 5 shows the analysis using It-Matrix as the dependent variable. Step 1 included Age at CI and Age at Last Follow Up and the model was entirely explained by Age at CI alone that accounted for almost 40% of the variance. In step 2, PCA-W and PCA-S were computed to the model, adding a further 21% to the overall variance, thereby reaching 61% of explained variance. In this second step the model variance was explained by PCA-S, while age at CI was excluded by the model. In step 3, TROG-2, which described morphosyntactic comprehension, was added and apportioned an additional 3.6%, for a total 65% of explainable variances. This model suggests that there is a specific contribution of PCA-S and TROG-2 to It-Matrix.

4. Discussion

Long-term assessments of implanted children are important inputs to guide families and professionals in therapeutic and counselling processes. There are relatively few studies reporting on long-term outcomes in adolescents and young adults implanted during childhood, and who have grown up with CI. Hence the present study aimed to assess the long-term speech and language outcomes in a sample of prelingually DHH adolescents and young adults.

4.1 Primary outcome: Long-term speech perception outcomes and morphosyntactic comprehension

Speech perception tests in quiet are being used routinely for the assessment of performance benefits in pediatric and adult CI users. CI users generally demonstrate higher level performance in quiet listening conditions. Although these test materials provide valuable information regarding users' performance, they do not give realistic information for adverse conditions such as listening in the presence of background noise at different SNRs [38]~~[38]~~. The present study provided, for the first time, the long-term outcomes for speech perception in quiet and in noise tests conducted in Italian, measured in a heterogeneous sample of adolescents and young adults implanted during childhood. One primary outcome of the present study was to evaluate long-term speech perception of words and sentences in quiet and with fixed SNR, and in adaptive noise using the Italian adaptation of the Oldenburg sentence test (It-Matrix). The data that emerged in our study showed that the use of CI positively contributed to the recognition of sentences in quiet and in noise, even in children with late access to surgery. However, the average values for speech perception in adaptive noise were significantly worse when compared to normative values, and only 9.2% of subjects fell within the normative range. Accordingly, words and sentence recognition at SNR + 10 dB and +5 dB showed a noteworthy performance deterioration compared to speech tests in quiet.

There is a paucity of papers reporting outcomes for fixed SNR and adaptive noise tests collected over the longer term. As mentioned above, the Uziel et al. study [19]~~[18]~~ involving 79 adolescents reported mean word recognition in quiet and with SNR+10 dB of 72% and 44% respectively. Age at CI was found to be a strong predictor of outcomes. Davidson et al. [20]~~[19]~~ performed a study observing mid- and long-term outcomes in a group of 112 teenagers. The authors observed that open-set recognition scores for words and sentences increased significantly as a function of increased age and listening experience: word recognition in quiet was 60% and sentences recognition in quiet and in noise were 80% and 52%, respectively. Outcomes recorded in the present study were only slightly better being 94% and 71% for words in quiet and with SNR +10 dB, and 89% and 64% for sentences in quiet and with SNR +10 dB. These differences might be explained by the different test materials, but also by a higher mean age at test and a longer follow-up for subjects belonging to the present

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study. As discussed by Davidson et al. [20][19] and Beadle et al. [39][39] speech perception and language skills tend to improve with age, positively influenced by increased experience with CI and by taking advantage of improved linguistic competence [40][40].

Regarding the current literature on It-Matrix sentence tests, there are currently two articles on adults and only one article on pediatric subjects. Gallo et al. [24][23] explored It-Matrix in a cohort of 45 unilateral and bimodal CI users, with a median age at test of 50 years and with short-term CI use. In their sample, unilateral and bimodal users achieved a median SRT of 4.15 dB and 2.85 dB respectively, with a wide range of outcomes. Dincer D'Alessandro et al.[25][24] assessed a group of 20 unilateral CI users with a median age of 65, and a median SRT of 7.6 dB SNR, once again with wide intrasubject variability. Concerning the pediatric population, Forli et al. [41][41] analyzed a sample of 36 children with bilateral sequential CI and a mean age of 11 years and a mean inter-implant time of 5 years. The mean SRTs reported were 3.9 dB with the first CI and 2 dB with the bilateral sequential CI respectively, which were found to be significantly different. Overall, the results from the above studies show large variability dependent on the sample examined (age, audiological characteristics, listening mode) and on the assessment setting.

It-Matrix intelligibility in our sample population showed a median SRT of -1.1 dB SNR for the whole sample and a value of -0.6 dB, -2 dB and -1.7 dB SNR for unilateral, bilateral sequential and bilateral simultaneous users respectively. These results are considerably better when compared to the studies of the adult population by Gallo et al. and Dincer D'Alessandro et al. [24,25][23,24] but also when compared to the study on younger subjects by Forli et al. [41][41].

Concerning better outcomes for It-Matrix observed in our study when compared to the adult hearing population, different variables might support such differences. From a perceptive point of view, speech perception through CI is influenced by frequency resolution which is poorer in CI when compared to hearing subjects [42][42]. Nevertheless, poor frequency resolution does not appear to limit speech perception in noise for prelingually deaf, early implanted children as much as it does for postlingually deaf adults. Short-term hearing deprivation and brain plasticity [43][43], which was observed in most children included in the present study, might have supported better outcomes as opposed to the adult population from Gallo et al.[24][23].

When comparing It-Matrix results to those found in the early implanted pediatric population by Forli et al.[41][41], better outcomes were observed for subjects included in the present study. Performance differences with worse outcomes reported by Forli et al. [41][41] might be due to younger mean age at test and the wider age range of subjects included in their work, whereas the standard It-Matrix test was administered in a younger pediatric sample (≥ 6 years). This once again is possibly related to the influence that age has on speech perception score, which increases significantly as a function of

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increased age and listening experience, being positively influenced by increased CI experience [20,39]~~[19,39]~~.

Children with different listening modes showed significantly better results for bilateral listeners in SF audiometric threshold, possibly owing to the summation effect measured in the current S0/N0 setting. Better SF thresholds increase audibility that in turn has been shown to be clinically more robust in bilateral when compared to unilateral and bimodal pediatric CI users, and in turn positively influence speech perception outcomes [44]~~[44]~~.

Sequential and simultaneous bilateral users did not show significant differences for speech perception in quiet and in noise. Most likely, such results reflect the fact that our sample included a high percentage of subjects with sequential implants (> 4 years) and there were no substantial age-related group differences at CI age. However, it may be argued that in the long term, as suggested by Kim et al. [45]~~[45]~~ the effects of the delayed sequential implantation might not be as relevant when speech discrimination is assessed in the S0/N0 testing mode. This last outcome once again might be linked to the prevalence of unilateral users in the study group and to the prevalence of subjects with sequential implantation in the bilateral subgroup. The composition of the sample size was determined by a more recent diffusion of the UNHS in Italy [46]~~[46]~~ and by differences in funding models between countries [47]~~[47]~~. Therefore, there are still many patients who received unilateral implant or delayed sequential bilateral implant in childhood who are now young adults. Finally, although group differences between unilateral and bilateral users (1.4 dB SNR) were not statistically significant at It-Matrix, differences higher than 1 dB SNR are considered clinically significant [48]~~[48].~~

Morphosyntactic comprehension is considered an independent contributor to open-set speech perception in paediatric HA or CI users [26,27]~~**[26,27]**~~**. Furthermore, morphosyntactic knowledge is fundamental when determining reading comprehension skills in DHH subjects and could be considered one of the most important factors that explain the variances observed in DHH students** [29]~~**[29]**~~**. The present sample showed wide inter-individual variability for this skill. Although the percentage of subjects within the normal range (a score ≥ 85) was 60%, the other 40% didn't achieve normal syntactic comprehension despite a mean period of 16 years of CI use. Indeed, children with higher scores in this kind of task have probably developed the skill set necessary to recognize entire sentences with the support of syntactic knowledge** [28]~~**[28]**~~**.**

During childhood, several studies highlighted the risk for CI users to show fragility in the domain of morphosyntactic skills, both in the expressive and comprehension areas [49–55]~~[25,49–54]~~. CI recipient children tend to reveal poorer grammatical processing such as omissions or substitutions of clitic pronouns marking gender or number, and verbal flexions [50]~~[49]~~. They also show poorer performance when using complex sentence structures and such outcome differences with hearing peers are still present after 5 years of CI use [51]~~[50]~~.

As for morphosyntactic production, variability in results was also reported in comprehension [52–55]~~[51–54]~~. Regarding this aspect, Geers et al. [54]~~[53]~~ found that about two thirds of CI recipient children aged 5–7 years and implanted within 5 years of chronological age, scored one standard deviation or more below controls on language comprehension. Schorr et al. [53]~~[52]~~ reported similar results for CI recipient children aged 5–14 years (age at implant 0;11 to 5;1), although this proportion decreases to 40% in Geers et al. [52]~~[51]~~, and down to one third in Hansson et al. [55]~~[54]~~.

The present study is, at this time, the only one that has been published concerning morphosyntactic comprehension of CI adolescents and young adults who were implanted during childhood. Therefore, it is not possible to make a direct comparison with other studies. If we compare our findings with the above reported literature, it suggests that the proportion of CI subjects with problems in morphosyntactic comprehension to be about 30%-40% as already indicated by Geers et al. and Hansson et al. [52,55]~~[51,54]~~, and this deficit can still be observed after 16 or more years of CI use and during adulthood. These results differ from those which were reported by Breland et al. [56]~~[55]~~, who found that differences detectable during earlier years of school may disappear during the high-school period. The present study included subjects who were implanted more than 20 years ago, when diagnosis of hearing loss and intervention was more delayed when compared to more recent years. Early intervention is one of the primary acknowledged factors impacting linguistic outcomes [57]~~[56]~~ and the present findings showed significant effects of age at implantation on morphosyntactic development.

Finally, statistically significant differences were not observed in our study even when dividing subjects into subgroups according to listening mode. Also, in this case, as discussed in the speech perception section, it could possibly be due to the smaller sample size and higher number of sequential versus simultaneous patients. In fact, other studies have shown better outcomes in bilateral young adult users with at least 10 years use regarding the development of receptive and expressive outcomes when compared to their peers with unilateral CI [58–60]~~[57–59]~~.

4.2 Secondary outcome: correlations between subjective, audiological variables and morphosyntactic comprehension

The bivariate analysis showed a significant correlation between speech perception and age at implantation especially in difficult listening conditions: the effects of age at CI persist into adulthood and both speech perception at fixed SNR and It-Matrix were inversely correlated with age at CI even when data were adjusted for age at test.

These results are in line with those described by Zalt et al. [22][21] in their long-term case control study. The authors found significant differences in outcomes for speech perception in quiet and for the Hebrew Matrix in all subgroups when compared to hearing controls. These results were mainly influenced by age at diagnosis and implantation despite a large subjective variability. Also, Pearson analysis showed a significant correlation between speech-in-noise and Raven, the receptive vocabulary, and the phonemic fluency score, underlying once again how speech perception is highly correlated to language.

Furthermore, our study highlights the correlation between the It-Matrix results and recognition of words and sentences in quiet and in fixed signal noise and this outcome is not influenced by age at CI or age at test. In general, It-Matrix results for the whole sample had great interpersonal and intergroup variability with similar results being found by Zaltz et al.[22][21] (SRT values between -4.5 and +1.25; a range of approximately 17 dB SNR). In addition, we demonstrated no differences between unilateral or bilateral samples.

The correlation of It-Matrix with speech perception in noise with fixed SNR +5 dB and +10 dB is a further novelty of the present study, as there is no other study which confirms this correlation both in the Italian language and for young CI users. Only a few studies have compared two or more different sentence-level SIN tests in the hearing-impaired adult population [61–63][60–62]. In particular, Jansen et al. [62][61] observed how the French Matrix had a significant correlation with everyday sentences in noise tests showing that the newly developed test was accurate and reliable in a large group of hearing and DHH subjects. In our opinion, the presence of a strong correlation between the standard It-Matrix and the Italian standard words and sentences in noise tests supports reliability and accuracy of measurements in the adolescent and young adult early implanted population.

Concerning morphosyntactic comprehension, this skill was found to be strongly correlated to It-Matrix and age at implantation. However, using a stepwise multiple regression analysis the weight of age at the time of implant decreased when predicting performance over It-Matrix and morphosyntactic comprehension. Late implantation age is therefore a risk factor, but it does not of itself explain the differences in auditory perception in noise or language development. Regarding the It-Matrix test, the most significant variables were PCA-S and TROG-2 (morphosyntactic comprehension). PCA-S alone explained 60% of variance and TROG-2 added 3% to the total variance. The ability to adequately perceive and repeat sentences, both in quiet and noise, was therefore the greater contributor to CI users' performance in listening with adaptive noise, probably due to the similarity between all of these tasks where the subject is asked to repeat a series of words, maintaining not only their semantic meaning, as happens for word recognition tasks, but also processing their role and function in sentence structure. This is an ability that, with respect to word recognition tasks, also requires more mature and efficient competences regarding other cognitive factors, such as working memory. In fact, in a study on postlingually deafened adult CI users, Kaandorp et al. [64]~~[63]~~ observed that the working memory capacity alone, measured through the Reading Span, explained 55% of the variance in SIN thresholds, suggesting that poor verbal working memory capacity limits speech-recognition abilities in CI listeners. In the same way, working memory may also influence performance in adults who grew up with CI and this facet should receive more attention.

Regarding the role of linguistic aspects, which, in the present study, were represented by the scores obtained at TROG-2. Various authors have already probed the independent contributions of speech production and language ability to open-set speech perception scores in children using either HAs or CIs [26,27]~~[26,27]~~. Furthermore, Eisenberg et al. [28]~~[28]~~ found that children's performance in Comprehensive Assessment of Spoken Language, a test structured to assess subjects' skills in syntactic constructions and paragraph comprehension, yielded the strongest correlation with sentence recognition. They concluded that children with higher scores in this type of syntactic task have probably developed, at the same time, the skill set necessary to recognize entire sentences with the support of syntactic knowledge and this phenomenon is already detectable commencing 5-6 years after CI activation. Our study supports the hypothesis that syntactic skills are still influential in adulthood, but with a smaller contribution than that measured by Eisenberg et al. in the pediatric population [28]~~[28]~~.

Limitations and future directions

A limitation of the present study concerns the paucity of bilateral simultaneous users in the study sample. Because of the small sample size and the absence of differences in age range at implantation between sequential and concurrent users, a comparative analysis was performed but was not statistically significant. However, it is possible that by supplementing the sample of simultaneous bilateral users with the results of the next generation of implanted children, these results may change, as nowadays simultaneous bilateral implantation is becoming an earlier and more widespread indication than in the past. Another limitation of the present study is the fact that, due to the retrospective nature of data collection, we were unable to test for other variables that could have explained the 40% residual variance. One of these variables is non-verbal intelligence, which has been found to be significantly correlated with Matrix outcomes in adolescent and young adult CI users by Zaltz et al. [22]~~[21]~~. Conversely, non-verbal intelligence was not found to correlate significantly with adaptive speech perception in noise outcomes in CI recipient children with typical and atypical language development by Torkildsen et al. [65]~~[64]~~.

Non-verbal intelligence has been found to correlate with receptive and expressive language skills in children with associated disabilities [14][13]. In contrast to the Meinzen-Derr study, non-verbal intelligence didn't reach statistical significance within a model of linear correlation including early CI and socioeconomic status. Particularly early implantation remains a dominant factor in children and adolescents, leading to better outcomes [66][65]. Similarly, the effects of non-verbal intelligence in a large cohort of early implanted children showed no significant correlations with postoperative comprehensive assessments of spoken language [67][66].

Differing results concerning the effects of non-verbal IQ between various studies might be as a result of different test methodologies and the lack of normative data for children with hearing loss, disadvantaging them in terms of clinical interpretation and recommendations [67][66].

The present study, being retrospective in nature, did not have complete non-verbal intelligence data, and therefore, could not contribute to shedding light on this important aspect.

Nevertheless, the outcomes of this study have provided a solid foundation for future prospective research, which should be designed to verify the role of variables such as auditory attention, working memory and cognitive functions on speech perception in noise and linguistic skills.

Conclusions

The results of the present study show the positive contribution made by CI in speech perception in quiet and in noise and in morphosyntactic skills, in DHH adolescents and young adults who received CI in childhood. Performances in quiet and in noise with fixed and adaptive SNR were, on average, better than that reported in the DHH adult population, despite wide variability in individual performances. Age at CI still represents an important factor influencing outcomes in long-term assessments. The study also provides insight into how speech perception and morphosyntactic comprehension are still interlinked in adolescence and adulthood.

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Table 1. Descriptive statistic of subgroups: mean and median values concerning subjective and audiological characteristics for unilateral and bilateral users. Bilateral users were further subgrouped into simultaneous and sequential. Median sequential inter-implant time was 9 years (range 3 – 11).

	Bilateral simultaneous (N=9)		Bilateral sequential (N=12)		Bilateral (N=21)		Unilateral users (N=33)	
Subjective variables	**Mean**	**SD**	**Mean**	**SD**	**Mean**	**SD**	**Mean**	**SD**
Age at diagnosis (months)	16.3	10	10.9	5.7	13.4	8.8	14.1	6.7
Age at HA (months)	18.2	10.2	12.5	5.8	15	8.8	15.6	7
Age at CI (months)	41.2	36.9	31.3	23.2	32.7	25	37.4	19.1
Age at last follow up (years)	16.3	5.8	22.1	2.4	19.1	4.7	18.5	3.7
Follow up length (years)	12.9	2.9	20	3.1	16.4	4.6	15.3	2.7
Audiological variables	**Mean**	**SD**	**Mean**	**SD**	**Mean**	**SD**	**Mean**	**SD**
SF 500-4000 Hz dB	29	3.2	26.5	5	27.6	5	33.7	6
W/Q %	93.8	11.1	97.7	4.6	96	8.1	95	8.7
W/SNR +10 %	73	7	72.1	13.9	74	11.4	73	18.1
W/SNR +5 %	41.2	24.2	44	14.5	43	19.1	45	21
S/Q %	87	24.6	94.3	9.7	90	17.6	90	16.8
S/SNR +10 %	60	30.5	78	21.8	69.3	27.7	62.5	27.7
S/SNR +5 %	25.7	33.8	22	14.6	23.5	24.3	28.7	25.1

	Bilateral simultaneous (N=9)			Bilateral sequential (N=12)			Bilateral (N=21)			Unilateral users (N=33)		
	Median	**Range**		**Median**	**Range**		**Median**	**Range**		**Median**	**Range**	
		min	**max**		**min**	**max**		**min**	**max**		**min**	**max**
It-Matrix dB SNR	-1.7	-2.9	20	-2	-5.4	18	-1,7	-5.4	20	-0.6	-6.8	20
TROG-2	101	61	117	89	83	112	89	61	117	95	55	119

SD, standard deviation; HA, hearing aid; CI, cochlear implant; SF, sound field; Hz, hertz; W/Q, Words in quiet; W/SNR, Words/Sound Noise Ratio; S/Q, Sentences in quiet; S/SNR, Sentences/Sound Noise Ratio; It-Matrix, Italian Matrix; TROG-2, Test for Reception of Grammar.

Table 2. Bivariate Spearman's analysis. Rho and p values calculated between age at CI and at last follow-up, audiological variables (speech perception in quiet, with SNR +10 and 5, and It-Matrix) and TROG-2 as language variables.

Variables (unit)		Age at CI (months) Rho (p)	It-Matrix (dB SNR) Rho (p)	TROG-2 (score) Rho (p)
Age at CI (months)		--	**0.5 (<0.001)**	**-0.6 (<0.001)**
Age at last follow up (years)		**0.5 (0.001)**	0.2 (0.100)	**-0.5 (0.001)**
Words (RAU)	**quiet**	-0.1 (0.200)	**-0.5 (<0.001)**	**0.5 (<0.001)**
	SNR+10	-0.1 (0.300)	**-0.2 (0.040)**	0.2 (0.100)
	SNR+5	-0.1 (0.4)	**-0.3 (0.008)**	**0.3 (0.01)**
Sentences (RAU)	**quiet**	**-0.3 (0.020)**	**-0.5 (<0.001)**	**0.6 (<0.001)**
	SNR+10	**-0.5(<0.001)**	**-0.5 (<0.001)**	**0.6 (<0.001)**
	SNR+5	-0.3 (0.020)	**-0.4 (0.003)**	**0.6 (<0.001)**
SF (dB HL)		0.2 (0.09)	0.1 (0.2)	-0.1 (0.3)
It-Matrix (dB SNR)		**0.5 (<0.001)**	--	**-0.5 (<0.001)**

CI, cochlear implant; SF, sound field; HL, hearing level; Rau, Rationalized Arcsine Units; SNR +10 and +5, Signal-to-Noise Ratio evaluated at +10 and +5; It-Matrix, Italian Matrix test; TROG-2, Test for Reception of Grammar. In bold significant differences for p<0.05

Table 3. Principal Component Analysis (PCA) loadings for words and sentences recognition components.

Components	Variance explained	Loadings
PCA-W (RAU)	70%	
Quiet		0.72
SNR+10		0.89
SNR+5		0.88
PCA-S (RAU)	77%	
Quiet		0.83
SNR+10		0.94
SNR+5		0.85

PCA words, PCA-W; PCA sentences, PCA_S; Rau, Rationalized Arcsine Units; SNR +10 and +5, Signal-to-Noise Ratio evaluated at +10 and +5

Table 4. Partial Bivariate Spearman's correlation. Rho and p values were calculated controlling for age at CI and for age at last follow up. It-Matrix adaptive noise outcomes remained significantly correlated to morphosyntactic comprehension (TROG-2), when controlling for age at CI and for age at last follow-up. Also, words and sentences in quiet and with SNR +10 and 5 remained correlated to It-Matrix. Finally, when controlling for age at last follow-up, all speech perception and morphosyntactic comprehension tests were correlated to age at CI.

Controlling for Age at CI	**Age at last follow up (years)**	**It-Matrix (dB SNR)**	**TROG-2 (score)**
	Rho (p)	**Rho (p)**	**Rho (p)**
Age at diagnosis (months)	-0.1 (0.400)	-0.03 (0.800)	0.05 (0.700)
Age at last follow up (years)	-	-0.3 (0.800)	-0.1 (0.500)
PCA words recognition	-0.1 (0.400)	**-0.3 (0.030)**	**0.4 (0.003)**
PCA sentences recognition	-0.2 (0.100)	**-0.3 (0.060)**	**0.5 (<0.001)**
SF (dB HL)	-0.1 (0.200)	0.05 (0.700)	**-0.1 (0.300)**
It-Matrix (dB SNR)	-0.03 (0.800)	-	**-0.3 (0.020)**
Controlling for Age at last follow up	**Age at CI (months)**	**It-Matrix (dB SNR)**	**TROG-2 (score)**
	Rho (p)	**Rho (p)**	**Rho (p)**
Age at diagnosis (months)	**0.4 (0.009)**	0.1 (0.300)	-0.2 (0.200)
Age at CI (months)	-	**0.4 (0.005)**	**-0.5 (0.001)**
PCA words recognition	-0.1 (0.500)	**-0.3 (0.030)**	**0.4 (0.006)**
PCA sentences recognition	**-0.3 (0.030)**	**-0.4 (0.007)**	**0.6 (<0.001)**
SF (dB HL)	0.1 (0.400)	0.1 (0.500)	**-0.2 (0.100)**
It-Matrix (dB SNR)	**0.4 (0.005)**	-	**-0.5 (<0.001)**

CI, cochlear implant; SF, sound field; HL, hearing level; PCA, Principal component analysis; It-Matrix, Italian Matrix test; TROG-2, Test for Reception of Grammar. In bold significant values for $p \leq 0.05$

Table 5. Hierarchical regression analysis for It-Matrix.

Variables	**STEP 1**	**STEP 2**	**STEP 3**
	β (p)	**β (p)**	**β (p)**
Age at CI	**0.630 (<0.001)**	0.153 (0.196)	0.011 (0.935)
Age at last follow up	-0.105 (0.406)	-0.091 (0.891)	-0.161 (0.157)
PCA words recognition		-0.007 (0.962)	-0.017 (0,921)
PCA sentences recognition		**-0.782 (<0.001)**	**-0.505 (0.002)**
TROG-2			**-0.352 (0.027)**
ΔR^2		0.214	0.036
R^2	0.397	0.611	0.647

PCA, Principal component analysis; It-Matrix, Italian Matrix test; TROG-2, Test for Reception of Grammar. In bold significant values for p≤0.05

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Declaration of interests

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.