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Long-term effectiveness of cochlear implants in prelingually deaf children

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Long-term effectiveness of cochlear implants in prelingually deaf children

Eficácia a longo prazo do implante coclear em crianças com surdez pré-linguagem

Trabalho final do 6ºano do Mestrado Integrado em Medicina com vista à atribuição do grau de Mestre

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ABSTRACT

Introduction: Cochlear implantation is a widely accepted method of auditory rehabilitation for individuals with severe to profound hearing impairment. Even though many studies have assessed the long-term results of the cochlear implant, very little use a follow-up period bigger than 10 years and there is still no consensus in the results stability over time. The aim of this investigation was to ascertain whether the auditory performance of prelingually deaf children changes, after a follow-up period of 15 years, using the cochlear implant.

Methods: From 1992 to 2001, 132 paediatric patients with severe to profound hearing loss received cochlear implants at the Department of Otorhinolaryngology of Centro Hospitalar e Universitário de Coimbra. Of these 132 children, 119 prelingually deaf children, who were followed for over 15 years after cochlear implantation, formed the study population of this retrospective chart review. A comparison between the results obtained 10 (T0) and 15 (T1) years after cochlear implantation, with the free field pure tone audiometry and language and speech perception tests, was performed.

Results: As for the free field pure tone audiometry, there was a statistically significant decrease in all the frequencies tested between T0 and T1. This decrease represents an improvement in the detection' sensibility of pure tones over the 5 years of follow-up.

As for the language and speech perception tests, there was a statistically significant improvement of the monosyllables and the numbers tests while, in the phrases and the 100 words tests, there was a statistically significant worsening of the results.

Discussion: The results observed in the monosyllables and the numbers tests could be related to a better auditory performance given by cochlear implants as well as by new speech processors. On the other hand, the results obtained with the phrases test may have decreased because the list of phrases used in the second assessment had a higher level of difficulty than the list used in the first assessment. Plus, a possible cause for the decrease in performance in the 100 words test may be related to the tiredness and lack of attention felt by children, as this test was always the last one to be done.

Conclusion*:* The results of this investigation revealed that, even after 15 years of use, the cochlear implant still offers, to patients with severe to profound hearing loss, an improvement in pure tones' recognition and an increase of the levels of speech perception and intelligibility.

KEYWORDS

Cochlear Implants | Prelingual Deafness | Child | Follow-up Studies | Speech Perception | Audiometry, Pure Tone

INTRODUCTION

Cochlear implantation is a widely accepted method of auditory rehabilitation for individuals with severe to profound hearing impairment, who cannot benefit from conventional hearing aids.[1,2]

The cochlear implant (CI) is an electrical device comprised by an external component (a microphone, a microprocessor and a transmitting coil) and an internal component (a receiver and an electrode array), which is inserted into the cochlea (**Figure 1**).

The main role of the external component is to extract intensity, frequency, and temporal cues from acoustic signals and to process them into an electrical code that is sent to the internal receiver. From there, this electrical signal is then delivered to the electrode array, which directly stimulates the remaining auditory nerve fibers in the cochlea.[1,3,4]

When looking at the advances in outcomes using the cochlear implant, it is important to value many aspects that lead to them. For example, the establishment of universal newborn hearing screenings was particularly important to increase identification of infant hearing loss and to allow earlier diagnosis and implantation at a younger age. In addition to this, the improvement of cochlear implant design and speech processing strategies provided even better results.[5]

Many studies have shown that cochlear implants have enabled children, not only to improve their speech perception but also their speech production, reading and writing skills, social development and academic outcomes.[6–8] At least, relatively to their delays in cognitive development. Maria Huber[9] conducted a study with 52 CI users with ages between 12 and 21 years old to evaluate their educational level, satisfaction with their vocational placement and correspondence between career aspiration and actual occupation. The study revealed that more than 80% of the school-aged children attended mainstream schools and that 12 out of 13 participants, who required work, were employed. However, the correspondence between their career aspiration and actual occupation was lower, compared to their normal-hearing peers.

Furthermore, Frederic Venail[10] found that from the 100 prelingually deaf children that formed his study population, 83% of the children aged between 8-11 years old and 67% of the children aged between 12-15 and without further disabilities attended mainstream schools. Additionally, from the participants older than 18 years, 50% had university-level education. This shows that implanted children ultimately achieve education levels similar to the general population and that they are adapting to the hearing world, which might be a reason why current studies ceased to compare implanted children with children using hearing aids and started comparing them to their normal hearing peers.[11]

Even though cochlear implants help in the development of oral language and speech perception, there is still considerable interindividual differences regarding their outcomes.[12–14] Factors such as age at diagnosis and implantation, aetiology of deafness, degree of preoperative hearing loss, cognitive ability, type of implant, duration of implant use, rehabilitation intervention approaches and family support, are possible causes for this variability.[13,15]

Depending on when the deafness occurred, we can divide it in prelingual, perilingual and post-lingual deafness. Prelingual deafness occurs before the acquisition of speech and language, perilingual occurs during this period and post-lingual occurs after. This distinction is important because each type of deafness will affect the auditory areas in its own way, depending on how much auditory stimulation there has been.

One of the events that can happen during auditory deprivation is crossmodal plasticity. Crossmodal plasticity occurs when cortical resources of a deprived sensory modality are recruited by an intact sensory modality. In this case, crossmodal plasticity occurs when the visual and somatosensory systems “take over” the auditory areas and recruit the processing resources normally used to process the auditory information.[16–19] This explains why auditory areas become active again despite of the lack of auditory stimulation.[20]

Lee[21] showed evidence of crossmodal plasticity using PET in a study of prelingually deaf children. He concluded that there is a positive correlation between low resting metabolic activity in the primary auditory cortex, prior to cochlear implantation, and post-implantation speech perception scores, suggesting that a lower recruitment of auditory resources by other sensory systems, leads to better outcomes in children with prelingual deafness.

Furthermore, Buckley and Tobey[19] showed that, when considering the influence of cross-modal plasticity on speech perception ability, age of onset of auditory deprivation seemed to be a more important variable than length of auditory deprivation. Their results suggested that crossmodal plasticity accounted for a bigger variability observed in speech perception performance in prelingually deaf children than in post-lingually deaf children.

Thus, further studies, regarding the long-term efficiency of cochlear implants, are required to evaluate whether crossmodal plasticity and other variables can influence these children’ auditory development and language acquisition over the years.

Even though many studies have assessed the long-term results of the CI, very little use a follow-up period bigger than 10 years and there is still no consensus in the results stability over time. Thus, it is essential to keep making long term follow-up studies, so it is easier to adapt rehabilitation plans to the needs of each child and to obtain better results with the CI.

The aim of this investigation is to ascertain whether the auditory performance of prelingually deaf children changes, after a follow-up period of 15 years, using the cochlear implant.

I hypothesize that the performance with the CI may still increase after 15 years of follow-up because, at this time, children may still be improving their auditory skills, due to the maturation of the auditory pathway.

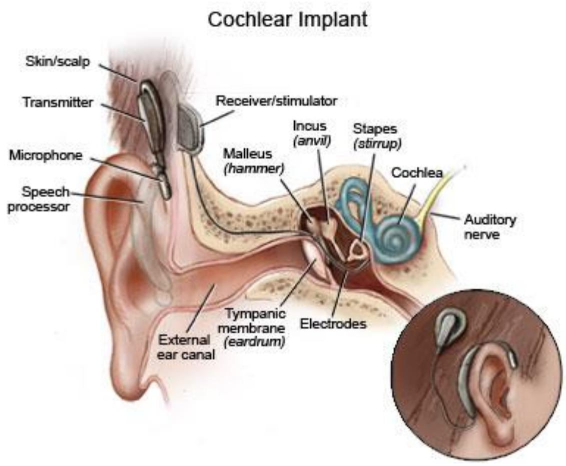

Figure 1 - Typical architecture of a modern cochlear implant (URL: https://www.nemours.org/patientfamily/khlibrary/articles/cochlear.html)

METHODS

Study Design and Participants

From 1992 to 2001, 132 paediatric patients with severe to profound hearing loss received cochlear implants at the Department of Otorhinolaryngology of Centro Hospitalar e Universitário de Coimbra (CHUC). Of these 132 children, 119 prelingually deaf children, who were followed at CHUC for over 15 years after implantation, formed the study population of this retrospective chart review.

Medical records were reviewed for age at implantation, duration of auditory deprivation, length of device use and cause of deafness (**Table 1**).

The average age of implantation was 3,14 ± 0,76 years old, with a minimum of 2 years old and a maximum of 5 years old and all of them were implanted unilaterally with Cochlear® models, mainly in the right ear (89,1%). None of the patients had hearing aids in the contralateral ear.

These patients were initially implanted with cochlear implants which had Cochlear™ Freedom® sound processors. However, many patients switched to Nucleus® CP810 or CP910 sound processors in the period between 10 and 15 years after implantation.

Cochlear implant use ranged between 19 and 29 years, with a mean of 22,2 years and a standard deviation (SD) of 2,7 years.

In most cases it was not possible to identify the cause of deafness (52%). However, 28% of the patients were found to have a genetical cause associated (mainly involving mutations in the Connexin 26 gene) and in 10% of the patients the deafness was caused by meningitis. Other causes such as cytomegalovirus and rubella infections, as well as traumatic lesions were associated with 10% of the cases.

Table 1 – Patients' Demographic

	M (SD)	Min-Max
Age	25,3 (3,03)	21-32
Age at implantation	3,14 (0,76)	2-5
Length of device use	22,2 (2,70)	19-29
	N	**%**
Aetiology		
Genetic	33	27,7
Meningitis	12	10,1
Others	12	10,1
Congenital unknown	62	52,1

Indications for cochlear implantation were according to the Portuguese guideline of Direção Geral da Saúde, regarding the treatment of deafness with cochlear implants in paediatric age.[22]

All implanted children were assessed daily during the first 1-3 months after implantation, depending on the age and the pre-implant language status of the child, then every 3 months during the first year and afterwards once a year.

The ethical approval to conduct this study was obtained from the Ethics Committee of the Faculty of Medicine, University of Coimbra. The Ethics Committee' process number for this study is "CE-122/2020", and it was approved on the 25th of November of 2020. All procedures were conducted following the Declaration of Helsinki.

Studied Variables

The audiological data collected included the free field pure tone audiometry and language and speech perception tests conducted 10 (T0) and 15 (T1) years after implantation. These are exams which are annually conducted within the usual medical care plan, without the need for additional specific tests.

The free field pure tone audiometry is a subjective test which identifies the hearing threshold levels of an individual by relying on its responses to pure tone stimuli. The pure-tone audiometry tests done in CHUC ranged between 250 hertz (Hz) and 6000 Hz.

The language and speech perception were assessed using the monosyllables test, the 100 words test, the numbers test and the phrases test.[23,24]

The Monosyllables Test is an open election test that evaluates the number of words and phonemes a patient can repeat from a combination of three lists with 20 monosyllables each, presented accordingly to age.

The Numbers Test is an open election test that evaluates the number of words and phonemes correctly repeated of a combination of two lists of numbers, presented accordingly to age.

The Phrases Test is an open election test that evaluates the number of keywords correctly repeated from a list of phrases, which contain highlighted keywords presented accordingly to age. Different lists of sentences were used, in the Department of Audiology and Speech Therapy, for children between the ages of 10 and 15 years (List 2) and for children older than 15 (List 3).

The 100 words test evaluates the number of correctly repeated words from a disyllabic word list presented according to age.

The language and speech perception tests were always performed in the same sequence. Firstly, the monosyllables test, followed by the numbers test, then the phrases test and lastly the 100 words test.

In all the tests above, a score of 0 was assigned when a child could not perform the task.

Statistical Analysis

The comparison between the audiological tests undertaken in T0 and T1 was made using the program SPSS – Statistical Programme for Social Sciences, version 26.0.

Firstly, it was assessed the normality of the samples using a Kolmogorov- Smirnov test. Since all the samples followed a non-normal distribution (with a significance level ≤ 0,05), it was used a non-parametric test, the Wilcoxon test, to compare the tests undertaken in T0 and T1. A value of $p \leq 0,05$ was considered statistically significant.

Descriptive statistics of the clinical variables (frequency, mean, standard deviation, minimum, maximum and percentiles) were also conducted.

Not all 119 participants were included simultaneously in the statistical analysis of each test because some of the participants had not completed them in T1 or had not done them in both time points. Thus, from the 119 participants, 116 were included in the analysis of the free field pure tone audiometry, 108 in the monosyllables test, 107 in the numbers test, 61 in the phrases test and 60 in the 100 words test (**Table 2 and 4**).

RESULTS

Descriptive statistics of the free field pure tone audiometry tests and of the speech perception tests undertaken in T0 and T1 are shown in **Table 2 and 4**, respectively.

As for the free field pure tone audiometry, the mean values obtained in all the frequencies (250 Hz – 6 kHz) decreased between T0 and T1 (**Figure 2 and Table 2**). In addition to this, the Wilcoxon test showed a statistically significant decrease in all the frequencies tested ($p \leq 0,05$). Z scores and *p*-values for each frequency are shown in **table 3**.

Table 2 – Differences between T0 and T1, for each audiometric frequency of the free field pure tone audiometry test

	N	Mean	(SD)	Min	Max	P25	P50 (median)	P75
250 Hz								
T0	116	34,35	(9,38)	15	55	30	35	40
T1	116	25,95	(9,09)	5	50	20	25	30
500 Hz								
T0	116	34,78	(7,90)	15	60	30	35	40
T1	116	29,22	(7,68)	10	50	25	30	35
1000 Hz								
T0	116	35,22	(7,44)	15	60	30	35	40
T1	116	29,22	(7,65)	10	50	25	30	35
2000 Hz								
T0	116	34,53	(7,34)	15	70	30	35	40
T1	116	30,04	(6,68)	10	45	25	30	35
4000 Hz								
T0	116	34,78	(6,54)	20	55	30	35	40
T1	116	31,90	(6,31)	10	55	30	30	35
6000 Hz								
T0	116	35,56	(7,80)	20	55	30	35	40
T1	116	31,77	(6,64)	10	50	26,3	30	35

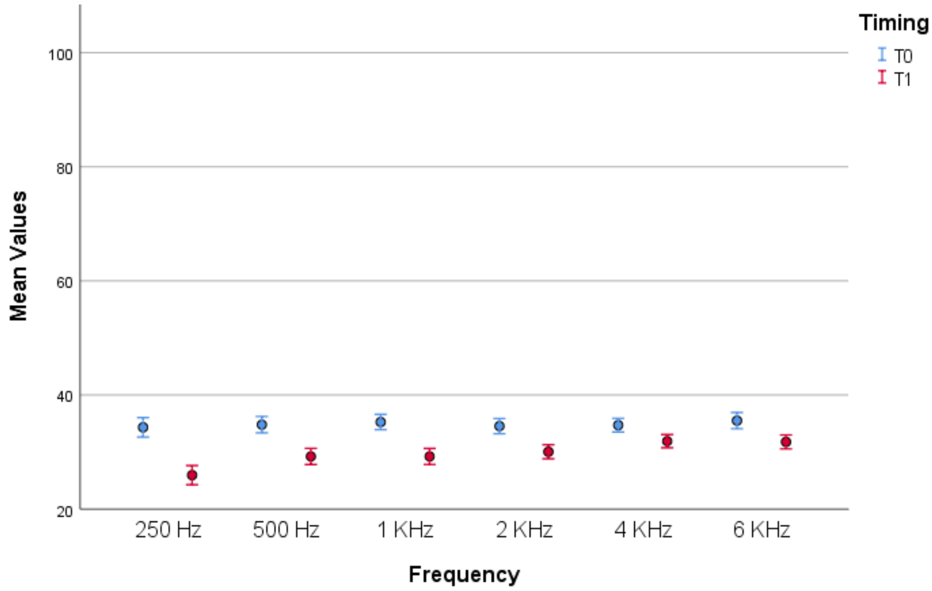

Figure 2 - Error Bar illustrating changes in mean between T0 and T1 within each audiometric frequency. Bars represent 95% confidence intervals.

Table 3 - Statistical analysis of the free field pure tone audiometry tests, using a Wilcoxon test.

	Z score	*p-value*
250 Hz	-6,26	**0,000**
500 Hz	-4,84	**0,000**
1 KHz	-4,96	**0,000**
2 KHz	-4,48	**0,000**
4 KHz	-3,59	**0,000**
6 KHz	-4,14	**0,000**

As shown in **figure 3**, mean values of the monosyllables test and of the numbers test increased between T0 and T1. In the monosyllables test, the mean score went from 60,46 to 73,06 and in the numbers test, it went from 89,12 to 92,20 (**Table 4**). This was supported by the Wilcoxon test, which showed a statistically significant improvement in the monosyllables test (z= -8,72, p= 0,00) and in the numbers test (z= -6,51, p= 0,00).

In contrast, the mean values of the phrases test and of the 100 words test decreased between T0 and T1. In the phrases test, the mean score went from 71,52 to 59 and in the 100 words test, it went from 86,57 to 69,18. Furthermore, the Wilcoxon test showed a statistically significant worsening in the phrases test (z= -4,73, p=0,00) and in the 100 words test (z= -6,57, p= 0,00).

Z scores and *p*-values for each test are shown in **Table 5**.

Table 4 – Differences between T0 and T1, for each language and speech perception test

	N	Mean	(SD)	Min	Max	P25	P50 (median)	P75
Monosyllables Test								
T0	108	60,46	(20,11)	0	88,3	48,3	65	76,6
T1	108	73,06	(20,73)	0	95,21	65,38	80,1	87,86
Numbers Test								
T0	107	89,12	(17,49)	0	100	90	93,3	100
T1	107	92,20	(16,46)	0	100	91,80	99	100
Phrases Test								
T0	61	71,52	(28,04)	0	100	52	84	93
T1	61	59	(31,73)	0	100	30	67	88
100 words test								
T0	60	86,57	(15,06)	33	100	82,50	92	96
T1	60	69,18	(24,49)	0	100	52,75	76	87,75

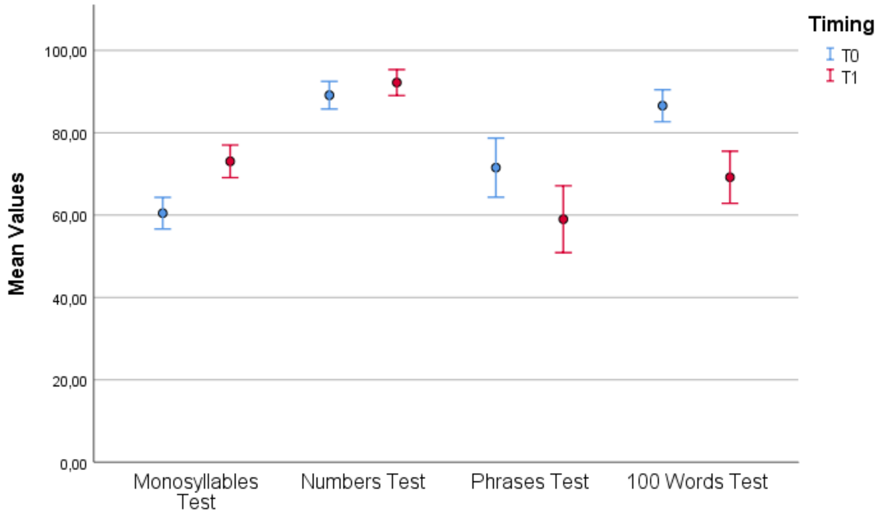

Figure 3 - Error Bar illustrating changes in mean between T0 and T1 for each language and speech perception test. Bars represent 95% confidence intervals.

Table 5 – Statistical analysis of the language and speech perception tests, using a Wilcoxon test.

	Z score	*p-value*
Monosyllables Test	-8,72	**0,000**
Numbers Test	-6,51	**0,000**
Phrases Test	-4,73	**0,000**
100 words test	-6,57	**0,000**

DISCUSSION

When comparing the results obtained with the free field pure tone audiometry at T0 and T1, there was a statistically significant difference among the audiometric frequencies obtained in the two assessments, with improvement in auditory sensibility at T1. This result represents an improvement in the detection' sensibility of pure tones over the 5 years of follow-up.

As for the language and speech perception tests, there was a statistically significant improvement of the monosyllables and the numbers tests, since the patients obtained better results after the 5 years of follow-up. The same did not happen with the phrases and the 100 words tests, as there was a statistically significant worsening of the results.

The improvement of the results registered in the monosyllables and the numbers tests, can be related to a better auditory performance given by cochlear implants as well as by new speech processors, which were installed during the period under investigation.

On the other hand, the worsening of the results in the phrases test may be related to an increase in difficulty of the tests. As said before, different lists of sentences were used for children between the ages of 10 and 15 years (List 2) and for children older than 15 (List 3). Therefore, the results may have worsened because the list of phrases used in the second assessment, List 3, has a higher level of difficulty than List 2, which was used in the first assessment.

In addition to this, a possible cause for the decrease in performance in the 100 words test is the fact that, this test, was always the last test to be executed by the patients. Therefore, the results could be influenced by the tiredness and lack of attention felt by these children, after executing so many tests.

A similar study was conducted in 2012, by Maria Peixoto,[23] using the same sample as this study but including more 3 prelingual, 3 perilingual and 7 post-lingual deaf children. The aim of her investigation was to compare the results obtained within the first year after cochlear implantation and the results obtained 10 years after implantation, using the free field pure tone audiometry test. Her study showed that the results were stable between the two assessments, except for the 2000 Hz frequency, which had an improvement of its results.

Therefore, it seems there has been a bigger improvement in the period between 10 and 15 years after implantation than in the first 10 years after implantation.

One reason that might explain this bigger improvement, is the technological advancement obtained with new speech processors, which are often installed within this timeframe.

Usually, when using a new processor, there can be a worsening of the auditory function in its first months of usage. This happens because, normally, the patients need a certain time to adapt to the new speech-coding strategies that these processors offer. However, in the long run, these next-generation sound processors end up improving these patients' auditory abilities, which might explain why the results were better after this period.

These findings are supported by Shu-Chen Peng's study,[25] who investigated the speech intelligibility levels of 24 prelingually deaf cochlear implant users with seven years of implant use. His results showed that children using cochlear implants with more advanced speech-coding strategies had better speech intelligibility scores.

On the other hand, there might be other reasons able to explain these findings. For example, there was a study conducted by Jiwani[26] which aimed to determine whether cortical auditory maturation was reached with long-term unilateral cochlear implant use. In his study, electrically-evoked cortical responses were recorded in 79 deaf children, who had short periods of bilateral auditory deprivation prior to implantation (2.03 ± 1.36 years), and in 58 normal hearing children. The study showed that the P1–N1–P2–N2 complex, which is the typical waveform of a mature auditory cortical response, began to emerge by 10 years of time-in-sound experience in both normal and deaf children. Moreover, it showed that the differences from the normal hearing waveform were bigger in deaf children who had less than 7 years of auditory experience with the cochlear implant while the differences became minimal after 15 years of auditory experience. Therefore, this study might explain why the results were better after 15 years of cochlear implant use than after 10 years.

Further studies describing the long-term effects of the cochlear implant after 15 years of cochlear implantation were not found.

With a follow-up period of 10 years, Waltzman,[27] showed an average rate of word recognition of 81% and a sentence recognition of 94%, in a study including 81 prelingually deafened children.

Also, Spencer,[28] in a study including 27 prelingually deaf children, presented a word and sentence recognition rate of 70% and 68%, respectively, during a mean follow-up period of 10 years.

Therefore, the scores obtained in the current study, after 10 years of follow-up, were slightly better in word recognition (86%) and slightly inferior in sentence recognition (71,52%), when comparing to the two studies above.

Even though these results represent a significant improvement in language and speech perception, there are other proven benefits of using cochlear implants, including achieving better academic results, more professional opportunities, more solid relationships and an overall improvement in quality of life.

For example, Elizabeth Beadle[29] assessed in her study, the auditory performance, speech intelligibility and academic status of a group of 30 profoundly deaf children. After 10 years of implant use, 87% of the children understood a conversation without lipreading, 60% used the phone with a familiar speaker and 77% had a speech intelligible to an average listener. From the 30 children included, 19 were in secondary schools (6 in mainstream schools, 7 were in specialist hearing-impaired units attached to the main secondary school and 6 were in schools for the deaf), 7 were in the university, 3 were working full-time and one was a full-time mother.

Furthermore, Alain Uziel[30] conducted a study with 82 prelingually deafened children to evaluate their speech perception and intelligibility and academic/occupational status. After 10 years of follow-up, mean scores of word recognition were 72%, 40% of the children had a speech intelligible to an average listener and 22% developed a speech intelligible to a listener with little experience of a deaf person's speech. Regarding educational status, 6 patients were at university, 3 were working, 14 were at high school and 32 were in junior high school.

Thus, not only it is important the hearing functional gain, but also the improvement of these patients' lives that comes with it.

There are many things that could be further investigated and improved in this study. For example, having a more detailed description of the patients would make it easier to associate certain variables to the results. For instance, knowing how many patients have had their processors updated would be important, so it could be verified whether it was truly influencing the results. Other information that would be important to know is the number of patients which had electrode failures during the period under investigation and the daily average use of the cochlear implant. Also, having more moments of auditory assessment during the 15 years of follow-up, would enable us to know more specifically how the results evolved during this period and detect whether there was a specific timing where the results started improving with the cochlear implant. Finally, it could have been used a bigger follow-up period, to further investigate the long-term effects of cochlear implantation.

CONCLUSION

Cochlear implantation has been offered at Centro Hospitalar e Universitário de Coimbra for more than three decades. For this study, 119 prelingually deaf children using cochlear implants were followed for a period of 15 years post-implantation. The results of this investigation revealed that, even after 15 years of use, the cochlear implant is still offering to these patients an improvement in pure tones' recognition and an increase of the levels of speech perception and intelligibility.

In a future study, it would be important to integrate the follow-up results of these patients over the next years, so it could be easier do detect further improvements or deteriorations of these patients' hearing capacity. Plus, it would also be relevant trying to explain the discrepancy of the results obtained with the different language and speech perception tests.

In conclusion, further studies are needed to help understand and address the persistent variability in outcomes when using cochlear implants.

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