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The French Cochlear Implant Registry (EPIIC): Cochlear implant candidacy assessment of off-label indications

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ABSTRACT

Objective:

Evaluate in France the outcomes of cochlear implantation outside the selection criteria, off-label.

Material and Methods:

This is a prospective cohort study including adults and children having received a cochlear implant (CI) in an off-label indication, that is outside the criteria established by the « Haute Autorité de Santé (HAS) » in 2012. The data was collected from the « EPIIC » registry on recipients who received CIs in France between 2011 and 2014. Speech audiometry was performed at 60 dB pre-implantation and after one year of CI use, as well as an evaluation of the scores of the quality of life with the APHAB questionnaire, the scores for CAP and the professional/academic status in pre and post implantation conditions. Major and minor complications at surgery have been recorded.

Results:

590 patients (447 adults and 143 children) with an off-label indication for CIs were included in this study from the EPIIC registry (11,7% of the whole cohort of EPIIC).

For adults, the median percentage of comprehension using monosyllabic word lists was 41% in pre-implantation condition versus 53% after one year of CI use ($p<0.001$) and 60% versus 71% in dissyllabic word lists ($p<0.001$). The CAP scores were 5 versus 6 in pre and post implantation conditions respectively ($p<0.001$) and the APHAB scores were statistically lower after implantation ($p<0.001$).

In the children cohort, the median percentage of comprehension using monosyllabic word lists was 51% in preimplantation condition and 65% after CI ($p<0.001$), and 48% versus 82% ($p<0.001$) for dissyllabic word lists. The CAP scores were 5 versus 7 respectively in pre and post CI conditions ($p<0.001$).

Thirty-two minor complications (5,4%) and 17 major complications (2,8%) were reported in our panel of off-label indication patients.

Conclusion:

These results suggest that a revision of the cochlear implantation candidacy criteria is necessary to allow more patients with severe or asymmetric hearing loss to benefit from a CI when there is an impact on quality of life despite the use of an optimal hearing aid.

Key words:

CI, outside the selection criteria, off-label indication, revision, speech audiometry, indications, single sided deafness, sensorineural deafness, partial deafness, hearing in noise.

INTRODUCTION

Cochlear Implant (CI) is now at a mature stage: the developments in terms of programming strategies, design improvements, especially the ones of electrode arrays, and the surgical techniques have progressively allowed the indication extension, for adults and children, with severe hearing loss and residual hearing, ensuring a high level of speech comprehension [1].

In France, the guideline for a CI referral centre is subject to the recommendations of the *Haute Autorité de Santé (HAS)* who, in 2012, specified the principal criteria to be observed when facing an adult or child candidacy for CI [2], criteria reminded in the recommendations for clinical practice in 2018 with the state of the art by the French Society of Otorhinolaryngology and Head and Neck Surgery [3].

However, since 2012, from the clinician's point of view, the selection criteria for CI candidacy have shifted. They evolved taking into account the phonetic material, measurement conditions, including hearing evaluation in noise condition [4] and quality of life questionnaires.

An increasing number of CI centres in France and abroad admit to have added off-limit selection criteria to the official ones ("off-label"). In France, CIs are indicated in bilateral severe to profound sensorineural deafness, with a 50% result for speech discrimination in quiet using Fournier word lists (or equivalent) at 60 dB with optimized hearing aids. By convention, the term off-label CI indication refers to any indication outside the usual criteria suggested by the HAS. They gather indications very close to the usual audiometric criteria (e.g., speech discrimination of 60% instead of the usual 50% at 60 dB) as well as indications that well overrun these criteria as, for example, single sided deafness with tinnitus. Thus, in a recent American study, over three quarters of the interviewed surgeons claim to carry out cochlear implantations outside the official criteria of the Food and Drug Administration (FDA) [5]. In this study, it seems to be a great disparity between centres in the matter of off-label indications, which leads to a great discrepancy regarding the unequal access to cochlear implantation through the territory. The literature data concerning these off-label indications is currently very poor [6-8].

The primary goal of this study is to have a situation analysis of CI off-label indications in France, using the EPIIC registry (Etude Post Inscription à la liste des Produits et Prestations Remboursables des systèmes d'Implant Cochléaire et du Tronc Cerebral) from its creation in November 2010 to February 2014.

MATERIAL AND METHODS

Patients

A prospective multicentric study was run from November 2010 to February 2014, and registered as "EPIIC". All the CI centres in France, certified by the ARS /ARH (Agences Régionales de Santé/Agences Régionales d'Hospitalisation) participated in this study.

All the patients that received at least one CI or brain stem implant between the 1st of January 2011 to the 28th of February 2014, and having given an oral consent for adults or parents

having given a written consent for the minors, were included in the EPIIC study. The exclusion criteria were the patients that had refused to participate to the EPIIC registry and the patients that were explanted and re-implanted within the same time frame who withdrew their consent.

Among the included patients of this study, only the patients with off-label indications were assessed. Off-label indication being the indication that do not meet the official HAS criteria.

Every patient underwent speech audiometry in free field before implantation, after a year of implantation with CI or ABI alone or associated with contralateral hearing aid. Evaluation of complications, quality of life with CAP scores, and APHAB questionnaires, as well as professional and academic achievement were collected before and after implantation.

The clinical data was collected in an anonymized file. Confidentiality conditions were approved by the Commission Nationale de l'Informatique et des Libertés (CNIL).

Audiometric speech assessment

Speech audiometry in free field at 60 dB was performed before implantation and one year after, implant only or with a contralateral hearing aid (HA). The test used monosyllabic and dissyllabic word lists.

Quality of life evaluation

CAP scale was assessed before and one year after implantation. This scale uses 7 different levels evaluating the auditive capacities of the patients.

The APHAB score was measured in adults. It consists of 24 items divided into 4 categories:

- Ease of communication
- Reverberation (communication reverberating environments)
- Background noise (communication in noisy environments)
- Sound aversion (intolerance to loud sounds)

The APHAB score reflected the percentage of difficulties experienced by the patient in the studied category, thus the lower the percentage, the better the results.

After implantation, professional and academic achievement were assessed.

Complications evaluation

The complications collected were split in two categories:

- Major complications: meningitis, revision surgery without replacement of the internal part (major scalp necrosis, severe infection, electrode array migration, eardrum perforation, repositioning of the implant, cholesteatoma, etc...), internal part replacement surgery, disabling tinnitus, facial nerve stimulation and pain not resolved with electrode deactivation and resulting in decreased benefit.

-Minor complications: transient facial paralysis, subcutaneous hematoma, infections not needing surgery, tinnitus, facial nerve stimulation, pain that can be solved by electrode deactivation and without benefit loss, non-persistent vertigo or dizziness.

Statistics

Statistical analysis used the Wilcoxon rank test for matched samples when the variable distribution was not normal and the Student test when it was normal. We used the median values in the Wilcoxon test and the mean values in the Student test. Logistic regression was performed to determine the patients gain predictive factors, based on speech audiometry scores using monosyllabic word lists pre-implantation.

In all cases, the test was considered suggestive for $p < 0.05$.

RESULTS

In total, 5051 patients were included in the EPIIC study.

590 (11.7%) patients were considered as being in an off-label indications with a mean age at first implantation of 46.3 (4.0-87.5) (Table 1).

Among these patients, 447 were adults (207 male, 240 female) and 143 children (69 male, 74 female) (table 1).

The CI indications for adults and children were progressive hearing loss in 57 patients, asymmetric hearing loss in 43 patients, disabling tinnitus in 11 patients, meningitis with onset of cochlear ossification in 9 patients, fluctuating hearing loss outside HAS criteria in 5 patients, contralateral neuroma in 8 patients, visual impairment in 8 patients, and no information given for 449 patients.

Speech audiometry in adult evaluation

150 patients underwent speech audiometry in free field with monosyllabic word lists before implantation and one year after with CI only condition.

The median percentage of comprehension in monosyllabic word lists was 41% before cochlear implantation and 53% after one year using CI only ($p > 0.05$) (Fig1).

105 patients underwent speech audiometry in free field with monosyllabic word lists before implantation and one year after with CI and contralateral HA.

The median percentage of comprehension in monosyllabic word lists was 41% before cochlear implantation and 74% using CI and contralateral HA ($p < 0.0001$) (Fig.1).

219 patients underwent speech audiometry in free field with dissyllabic word lists before implantation and one year after with implant only.

The median percentage of comprehension in dissyllabic word lists was 60% before cochlear implantation and 75% after one year of CI using CI only ($p < 0.0001$) (Fig.1).

160 patients underwent speech audiometry in free field with dissyllabic word lists before implantation and one year after with CI and contralateral HA.

The median percentage of comprehension in dissyllabic word lists was 60% before cochlear implantation and 90% using CI and contralateral HA ($p < 0.0001$) (Fig.1).

Quality of life evaluation in adults

270 patients had a CAP evaluation before cochlear implantation and one year after. The median of the CAP scores was 5 before implantation and 6 after ($p < 0.0001$ 5.3×10^{-14}) (Fig.2).

Mean percentage of communication difficulties, evaluated by APHAB scores, was 53.4% pre- and 30.5% post-implantation ($p < 0.0001$). The percentage of difficulties in reverberant conditions was on average 69.4% pre- and 56.3% post-implantation ($p < 0.0001$), and in background noise conditions scores were on average 73.4% before and 57.1% after implantation ($p < 0.0001$). Sound aversion was 46.7% pre- and 41.4% post-implantation ($p < 0.005$) (Fig.2).

Patients' improvement (percentage of comprehension gain) after cochlear implantation was obtained when the speech audiometry in monosyllabic word lists before implantation showed comprehension lower than 54.3% (Air Under the Curve 0.875) (Fig.3). Above this threshold, there was no improvement with CI.

Speech audiometry in children evaluation

34 patients underwent speech audiometry in free field with monosyllabic word lists before implantation and one year after with implant only.

The median comprehension percentage was 51% before CI and 65% one year after with CI only ($p < 0.005$) (Fig.4).

35 patients underwent speech audiometry in free field with monosyllabic word lists before implantation and one year after with CI and contralateral HA.

The median percentage of comprehension for speech audiometry in free field with monosyllabic word lists was 48% before implantation and 82% using CI and contralateral HA ($p < 0.0001$) (Fig.4).

86 patients underwent free field speech audiometry with dissyllabic word lists before implantation and one year after with CI only.

The median comprehension percentage was 80% before and after one year of CI use ($p > 0.05$) (Fig.4).

75 patients underwent free field audiometry in dissyllabic word lists before CI and one year after implantation with CI and contralateral HA conditions.

The median comprehension percentage in free field speech audiometry in dissyllabic word lists was 80% before CI and 90% using CI and contralateral HA ($p < 0.0001$) (Fig.4).

Quality of life evaluation in children

40 patients had a CAP evaluation before CI and one year after. The median CAP score was 5 before and 7 after CI ($p < 0.05$) (Fig.5).

Complications

7 patients (1.2%), children and adults, had intraoperative complications: 3 patients had a cerebrospinal fluid leak (0.5%), 1 patient a major bleeding (0.2%), 1 patient neurologic disorders (0.2%), 1 patient neurovegetative disorders (0.2%) and 1 patient for whom we have no information (0.2%).

In total, 49 patients (8.3%), children and adults, had postoperative complications, 32 minor complications (5.4%) and 17 major complications (2.9%): 5 skin infections (0.8%), 6 skin disorders (1%), 4 subcutaneous haematomas (0.7%), 3 facial paresis (0.5%), 14 dizziness (2.4%), 1 disabling pain (0.2%), 1 electrode array migration (0.2%), 3 tympanic perforations (0.5%), 3 disabling tinnitus (0.5%), 1 incorrect electrode placement (0.2%), 5 electrode or internal implant displacement (0.8%), and 3 implant failures (0.5%).

DISCUSSION

In this study on the EPIIC registry, the results for adults and children show an improvement of the speech audiometry and quality of life scores after CI when the indication was off-label.

Taking into account changes in indications to be supported financially by HAS requires modifications after marketing approval from industrials with sufficient level of safety proof, and good functional results certified by clinical studies. However, these studies are expensive and last several years, preventing access to CI for patients currently off-label, in particular the paediatric population for whom hearing deprivation must be as short as possible.

Nevertheless, there are many medical examples of the use of medical products outside the official indications allowed on the market. In ENT, the use of gentamicin, for example, to perform a chemical labyrinthectomy in the Meniere disease. The use of off-label therapies is justified by the physician when there is no alternative therapy available or when the positive results of the proposed therapy (e.g. CI) exceed those of available standard treatments. It is therefore decisive that clinical teams in CI centres and industrials collect as much data and results as possible to help new indications and recommendations to emerge and have them approved by health authorities. There should be, in particular, evaluation of the CI contribution in patients having a less severe hearing loss, as well as the benefit of bilateral implantation in off-label conditions.

Cochlear implantation and off-label hearing threshold

Defining a new audiometric threshold from which a CI would be better than a HA is difficult once the current recommended threshold is exceeded, 50% speech discrimination in quiet with the Fournier list (or equivalent) at 60 dB, in free field, with optimal HA, as well as the benefit of a bilateral implantation in different conditions than the ones recommended. This threshold value varies according to the hearing task, the frequency band and the performance goal set. A residual hearing better than 60 dB HL between 125 and 500 Hz would be required to reach the normal level of intonation perception according to Marx and al. [9], whereas the musical appreciation would be facilitated in bimodal stimulation if the residual hearing is better than 80 dB HL between 125 and 1000 Hz [10].

Five main predictive factors affecting the quality of speech perception post implantation are widely accepted in literature: aetiology, age at onset of severe to profound deafness, age at implantation, and duration of CI use [11].

However, the relationship between pre and post implantation speech perception scores has been little reported, mainly because of the methodological difficulties to carry out such studies and the difficulty to compare them depending on the language and the phonetic material used. The common pitfall found is the low proportion of patients having a pre implantation monosyllabic speech perception score well over 0%. Overall, the literature analysis shows that the post implantation speech scores are positively linked to the pre implantation speech scores, i.e. the better the pre implantation score, better the performance with the CI [1,7,12].

Preoperative speech assessment enables to determine the level of intelligibility at the most comfortable level, set at 60 dB in France, which can be close to discomfort threshold. The maximum intelligibility score, usually not taken into account in the CI selection criteria, is a measure of the HA performance-intensity relation, individual dependent, especially for greater hearing loss levels, which can edge or equal the discomfort level. The functional dynamic being reduced, the HA performance at everyday level is limited. The result is then a patient dissatisfaction and sometimes abandon of the HA. For most of the CI candidates, the maximal intelligibility score is higher than speech score at 60 dB [13, 14], reflecting a functional dynamic information in relation to the value at 60 dB. In a recent study from Hoppe and al. [15], it has been demonstrated in a panel of 284 implanted adults that a high score of maximum intelligibility preoperatively is predictive of the postoperative results, with 96% reliability, and that the speech intelligibility scores at 65 dB with the CI were higher than preoperatively at the intensity level for maximum speech intelligibility. Predictability of the postoperative results and patient counselling are paramount especially when the preoperative residual hearing level is high, as this residual hearing may disappear after cochlear implantation. A selection criteria extension has positive long-term effects on the speech performances and improves the quality of life of patients [16]. A rise of threshold from 50% to 60% of speech comprehension at 60 dB could bring a consensus in France, in both the clinical teams and the CI community.

CI and residual hearing

CI selection criteria have changed over time. Our data shows a great disparity in the speech assessment methods pre and post implantation: the dissyllabic word lists are widely used but they could underestimate the patient's unease and delay cochlear implantation, contrary to the use of monosyllabic word lists. Moreover, the intra-individual variability according to the type of speech evaluation is important and the clinical signification is of high importance to take the implantation decision [17].

Residual hearing or partial deafness is defined by a normal or mild to moderate hearing loss at low and medium frequencies, and a profound hearing loss at high frequencies, and other quantifiable hearing damage [3]. Cochlear implantation in patients with residual hearing has

been possible since a few years due to evolution of surgical techniques and technical developments (Electric acoustic stimulation implantation [EAS]) [18, 19].

The goal is residual hearing preservation at low frequencies after implantation. This approach allows electrical stimulation in the implanted ear through the CI in the missing high frequencies combined to an acoustic stimulation in the postoperative residual hearing.

As low frequencies are involved in the understanding of speech and music, patients retain a certain degree of capacity to detect vowels and the laryngeal fundamental (F0), but a very limited capacity to detect consonants (formant, spectral envelope), leading to comprehension difficulties in noise, even with HA. Patients with ski slope hearing loss, with over 30 dB/octave loss, show an impossible improvement of speech comprehension above 55 dB [20]. The functional benefits of bimodal stimulation on the same ear, acoustic and electric, have been the topic of many clinical investigations [21-27]. The advantages of a combined stimulation are a better speech discrimination in quiet and in noise and a better musical perception [19, 28].

These developments have enabled to extend official CI selection criteria to patients often outside the usual criteria because of their residual hearing, but who are uncomfortable even with bilateral HAs.

In this way, the group HEARRING suggests that the case of residual hearing is a potential indication for electro-acoustic cochlear implantation [29]. A hearing assessment in noise condition is required to identify the hearing impairment of these patients that reach 50% of speech comprehension in quiet due to their residual hearing [30].

Cochlear implantation and single sided deafness

Single sided deafness would affect 1.5% of the adult population [31] and CI is not currently recommended for these patients. The literature data shows effectiveness of electrical stimulation on unilateral tinnitus non responding to usual treatments and having devastating effects on the patient's quality of life [32-36]. The point in cochlear implantation in single sided deafness towards comprehension difficulties in noise and sound localisation seems to be more controversial, probably due to the current limitations of the CI ability to code the fine temporal structure of the acoustic signal [37, 38]. The controversial results on the sound source localisation would be related to technological limitations in communication between the implant and the contralateral HA for acoustic cues of interaural difference [39]. The duration of deafness on the totally deaf side also has a negative effect on the unilateral CI results [40], which raises the question of the importance of clinical context on the potential implant indication. New perceptive tests could also help to determine indications closer to the patient's everyday needs than usual tests, such as a test with moving sounds [41].

Few studies concern the implanted paediatric population with congenital or acquired single sided deafness. The results seem to be encouraging concerning speech comprehension in noise [42-46] with however a risk of CI abandon [47]. Implantation could also treat tinnitus due to unilateral deafness [44]. The results could be, as in adults, linked to the duration of deafness [33] although Sharma and al. have suggested a benefit of late cochlear implantation

even because of great cortical plasticity in childhood [48]. The expansion of hearing screening at birth has enabled an earlier diagnosis of congenital hearing loss, among them unilateral and progressive hearing loss, and early care of these pathologies that can affect language development, hence the importance of research to be done in the implantation domain concerning single sided deafness in paediatric population.

Cochlear implantation and vestibular schwannoma (VS)

The natural history of VS exposes to sudden deafness or progressive sensorineural deafness. Hearing preservation in VS surgery is restricted to total deafness or postoperative hearing loss of about 90% [49].

The hearing recovery in VS can be successfully expected through CI and has been suggested since 1995 [50]. Different situations can allow CI:

-VS with no surgery:

-CI on the side of the tumour, after observing a long period of stability [51].

-CI on the opposite side of the tumour which can be monitored by MRI, thanks to the compatibility now at 1.5 or 3 Tesla with diametric magnets (Med-El, Innsbruck; Advanced Bionics Corp.) not needing any contention during the process and minimizing the artefact by alinement of the magnet in the magnetic field axis.

-After stereotactic radiotherapy by Gammaknife [52].

-VS with simultaneous surgery (implantation after of tumoral removal) [53] or sequential surgery (remotely from the tumoral surgery) [54], especially after extralabyrinthic surgery (retrosigmoid, retrolabyrinthic or in the midbrain fossa) where hearing preservation has not been obtained postoperatively.

The sequential implantation requires an MRI evaluation of the labyrinthine liquids. In fact, a substantial change of the labyrinthine liquid signal has been seen in 1/3 of the cases in patients that had retrosigmoid surgery [55].

Several studies have showed the benefit of CI in patients suffering of sporadic VS on the best ear, with a functional prognosis close to a population of CI users without VS [56-59].

However, the choice of the surgical approach has an impact on the CI results: a translabyrinthic approach is related with poorer results [60]. When the contralateral hearing is normal, the benefit of CI on the operated VS side has recently been evaluated and seems to show benefits by restoring binaurality [61,62], and by decreasing tinnitus on the tumour side [62]. More recently, a CI has been suggested in a cochleoectomy of an intralabyrinthic schwannoma surgery with a good functional hearing outcome in 9 out of 10 cases [63].

Other pathologies than VS such as advanced otosclerosis and chronic labyrinthine otitis with a “weak” contralateral ear can be considered for cochlear implantation.

CONCLUSION

The national registry analysis shows variability in the off-label candidates for CI, due to the iniquity of the care propositions across the country. The significant percentage of referral centres performing cochlear implantation with off-label indications reflects the too restrictive and dated nature of the official recommendations. It demonstrates the progress made in a few years concerning the surgical techniques and the postoperative rehabilitation care, as well as the improvement of the processors. Our results show the CI benefit in a patients group having better preoperative speech comprehension skills than the official recommendations limit. Residual hearing requires systematic audiometric tests in noise and must be the indication for an electroacoustic implant. The clinician must be able to offer off-label cochlear implantation as soon as there is no therapeutic alternative or when the therapeutic results exceed those of existing treatments. The national registry has enabled to assess the CI benefit in patients with less severe hearing loss, as well as the benefit of bilateral CI in off-label situations (bacterial meningitis, temporal bone fracture and other causes of deafness that could lead to short term bilateral cochlear ossification). In 2018, the French Society of ORL and of Head and Neck Surgery, and the National College of Hearing Aid Specialists have recommended (n° 5, 11, 17, 23 and 24) evolution of selection criteria.

We suggest a complete update of official recommendations defining the cochlear implantation audiologic criteria to determine what is possible and acceptable to authorize larger indications, in order to allow a larger target population to benefit from the CI current performances.

Conflict of interest: none

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Fig. 1: Speech audiometry in free field at 60 dB, with monosyllabic and disyllabic word lists, of adult panel, before cochlear implantation, one year after implantation with implant only and one year after with implant and contralateral HA.

Fig. 2: Quality of life scores with CAP and APHAB before and after cochlear implantation in adult population.

Fig. 3: Audiometric gain assessment according to speech audiometry in free field at 60 dB with monosyllabic word lists, of adult panel, before cochlear implantation, one year after implantation with implant only or one year after with implant and contralateral HA. Patients with no improvement (red), patients with improvement (blue).

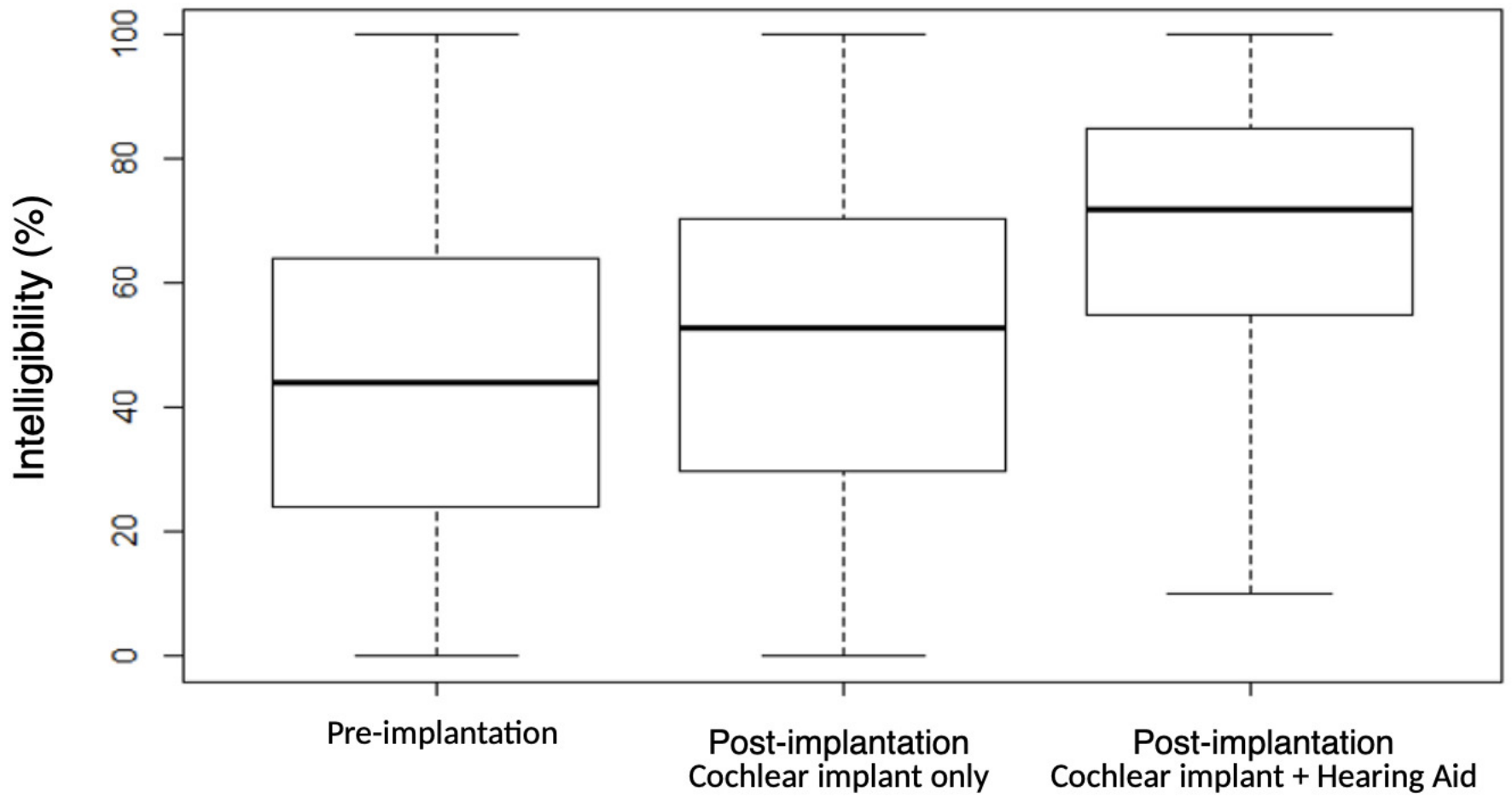
Fig. 4: Speech audiometry in free field at 60 dB, with monosyllabic and disyllabic word lists, of paediatric panel, before cochlear implantation, one year after implantation with implant only and one year after with implant and contralateral HA.

Fig. 5: CAP quality of life scores, before and after cochlear implantation, in paediatric population.

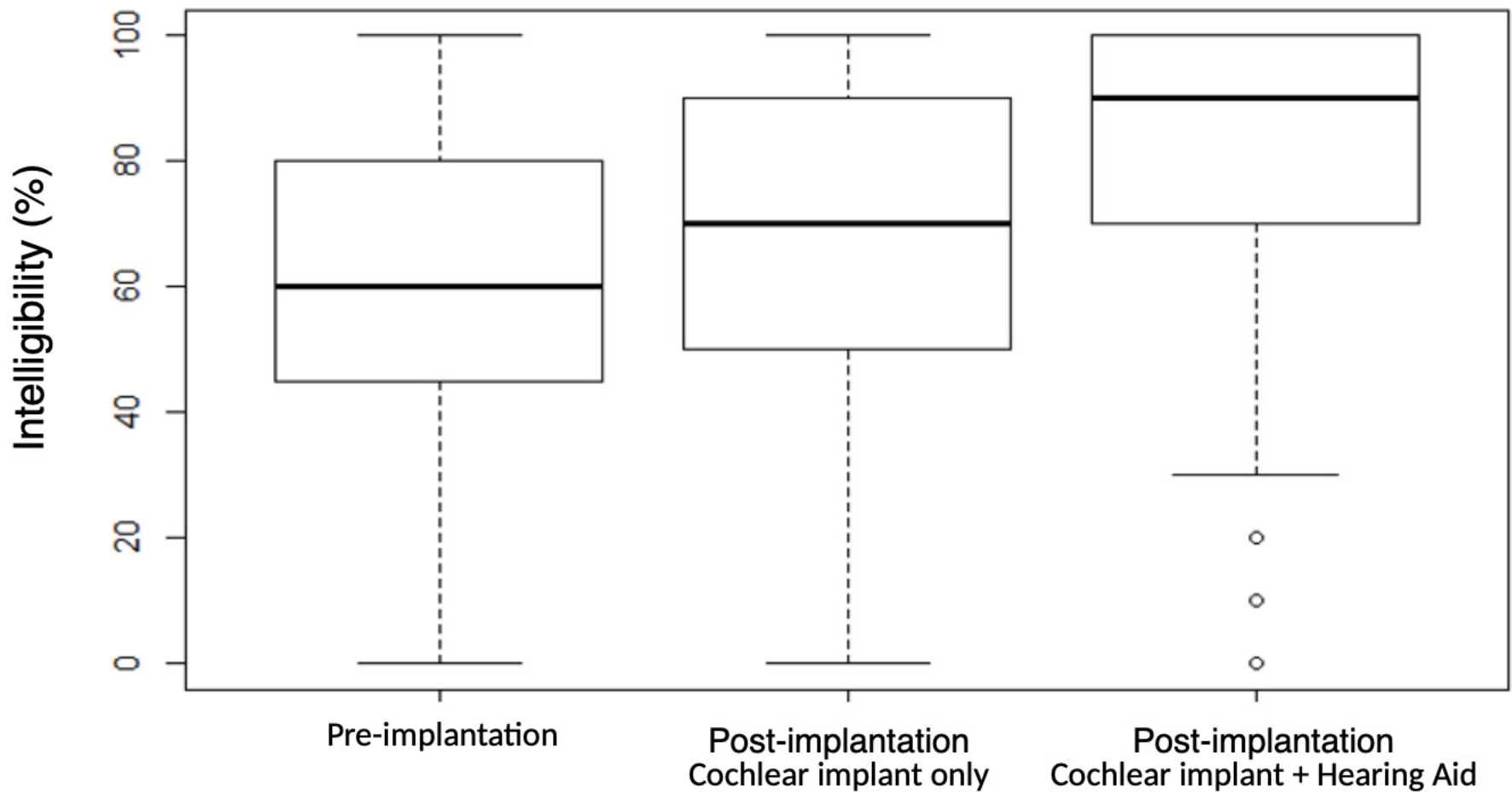
Gender	Male	207
	Female	240
	Children	143
Age	≤ 2 years	0
	2 years < to ≤ 6 years	31
	6 years < to ≤ 18 years	112
	18 years < to ≤ 35 years	52
	35 years < to ≤ 65 years	219
	> 65 years	176
Adults etiologies	Meningitis	6
	Cochlear Malformation	6
	Otosclerosis	45
	Others	192
	Unknown	175
	Undetermined	23
Children etiologies	Meningitis	3
	Cochlear Malformation	6
	Cochlear nerve hypoplasia	1
	Others	57
	Unknown	72
	Undetermined	4
Adults profession 1 year after cochlear implantation	Active	37
	Students	4
	Inactive	4
	Invalid	9
	Looking for work	17
	Retired	201
	Undetermined	175
Children scholarship before implantation versus 1 year after cochlear implantation	Special schools	15 vs 13
	Inclusion	59 vs 33
	Integration	62 vs 51
	Out of school	1 vs 0
	Undetermined	6 vs 45

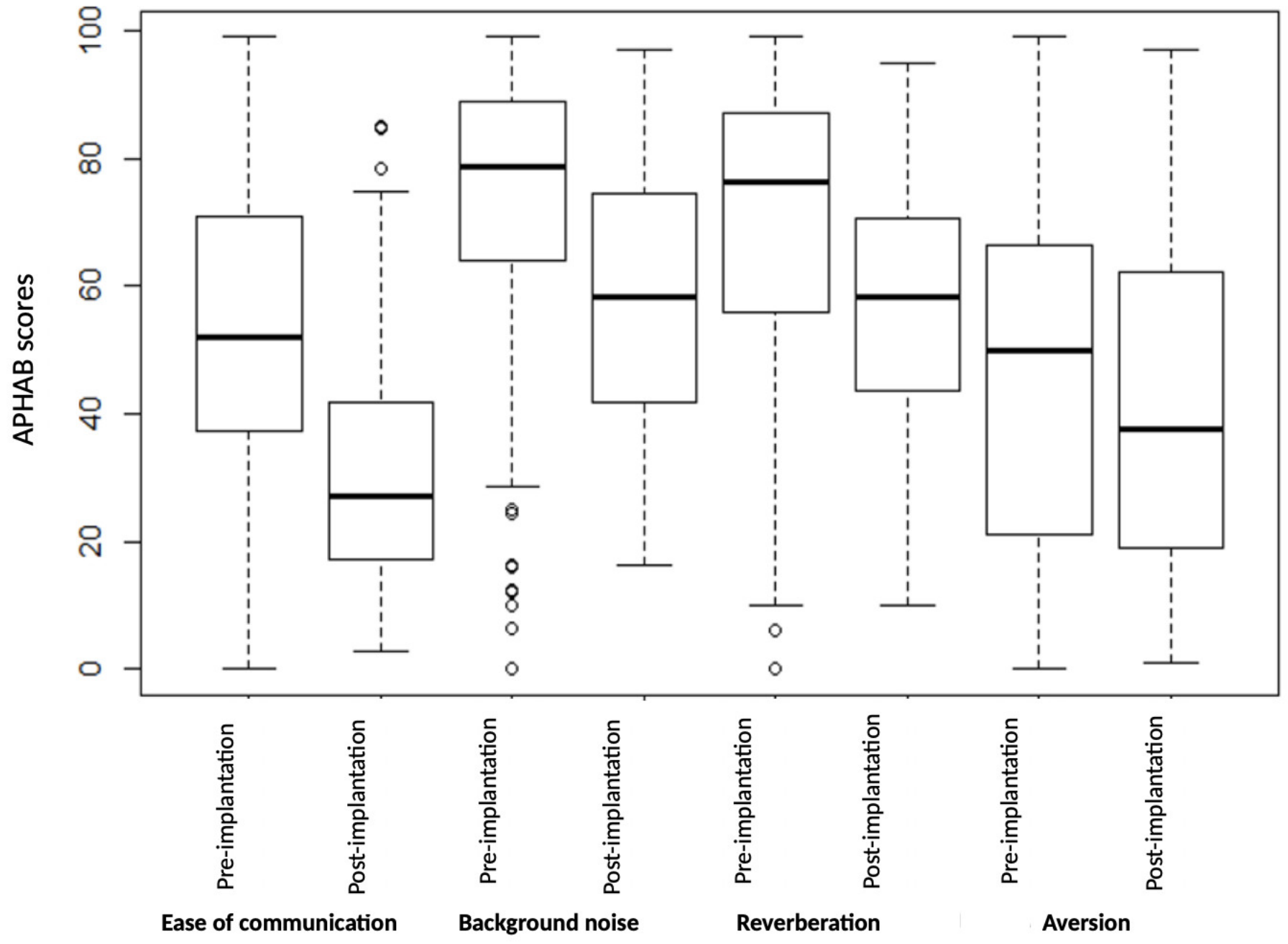
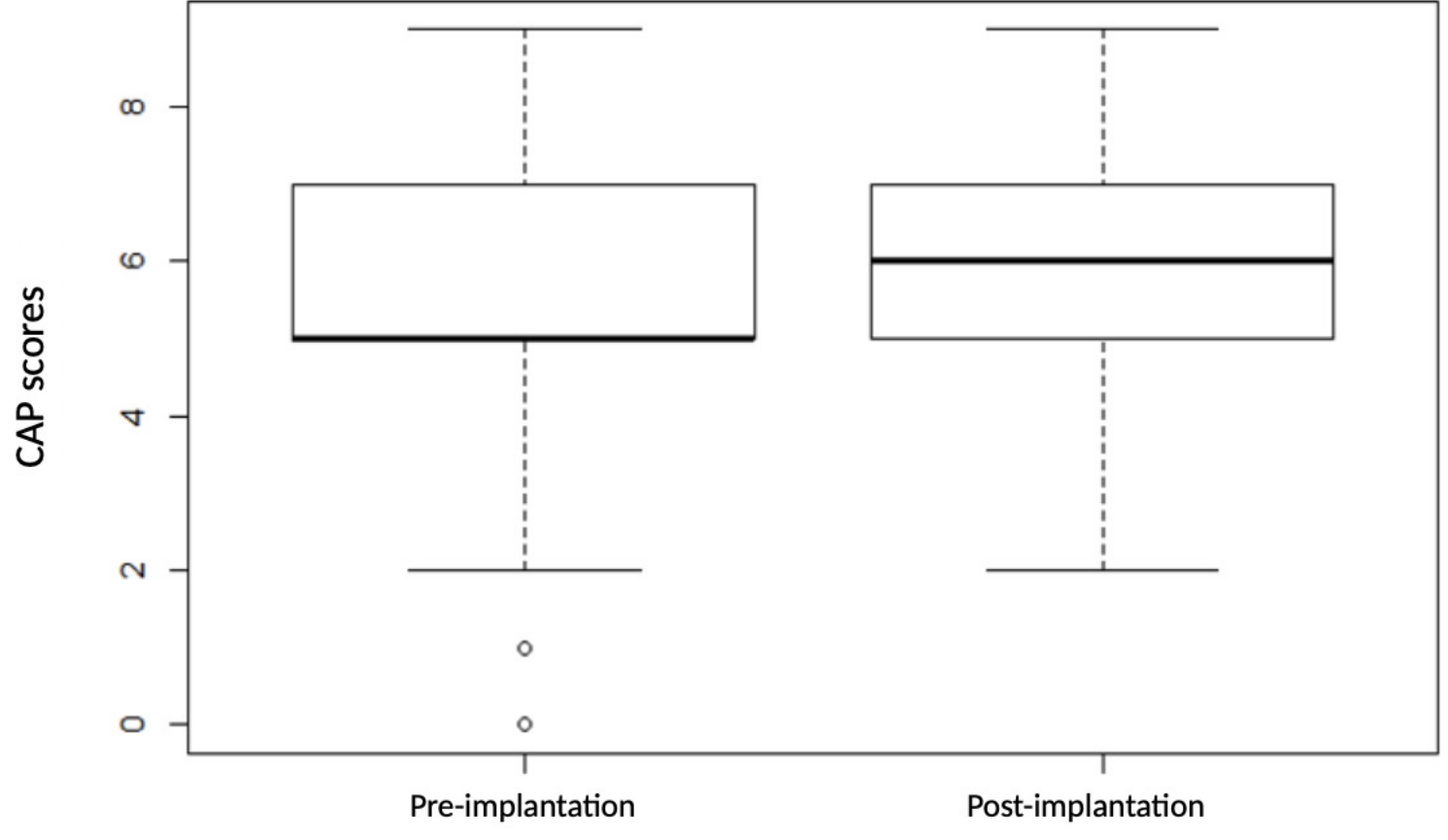
Table 1: Demographic data of patients in CI off-label indications.

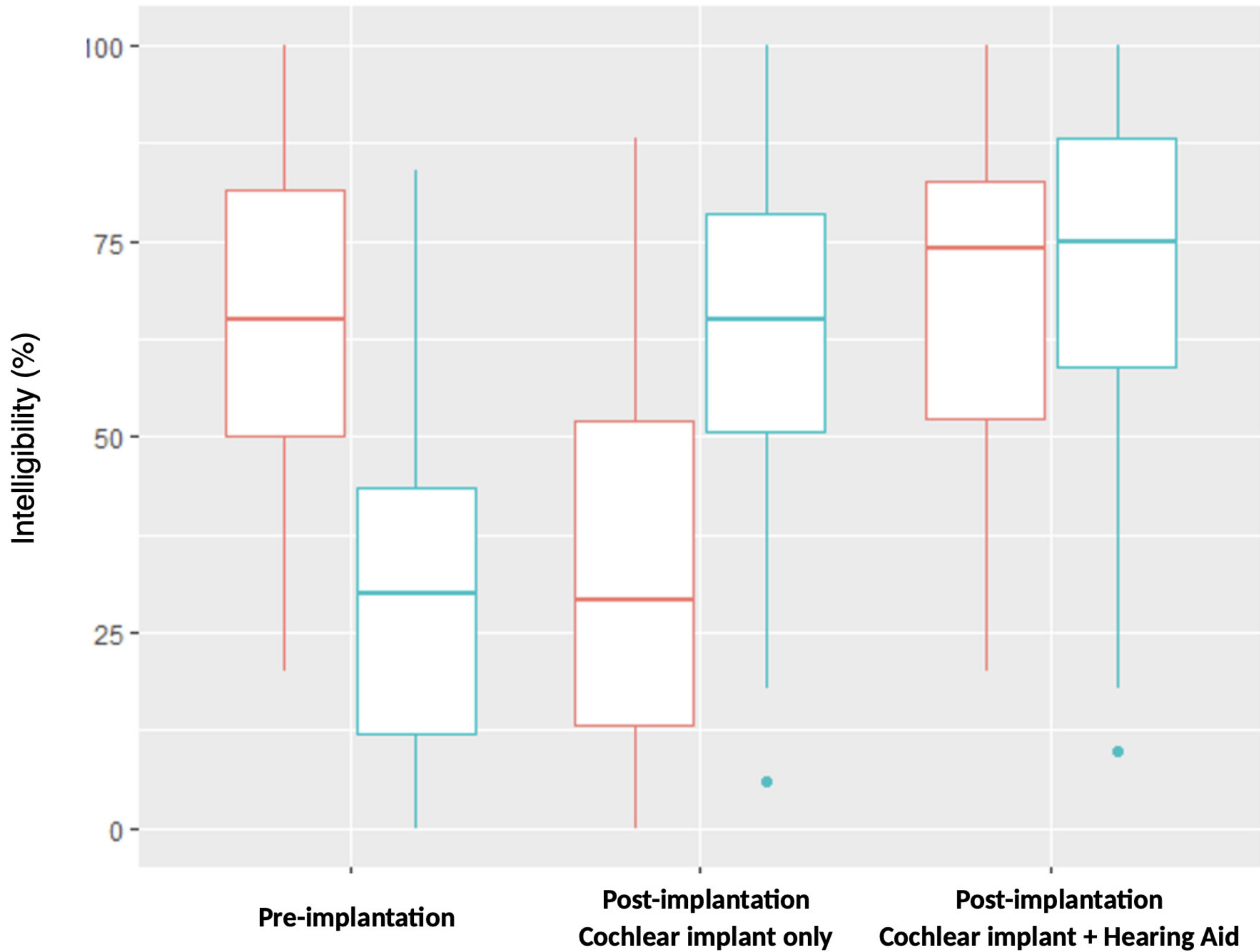
Monosyllabic word lists



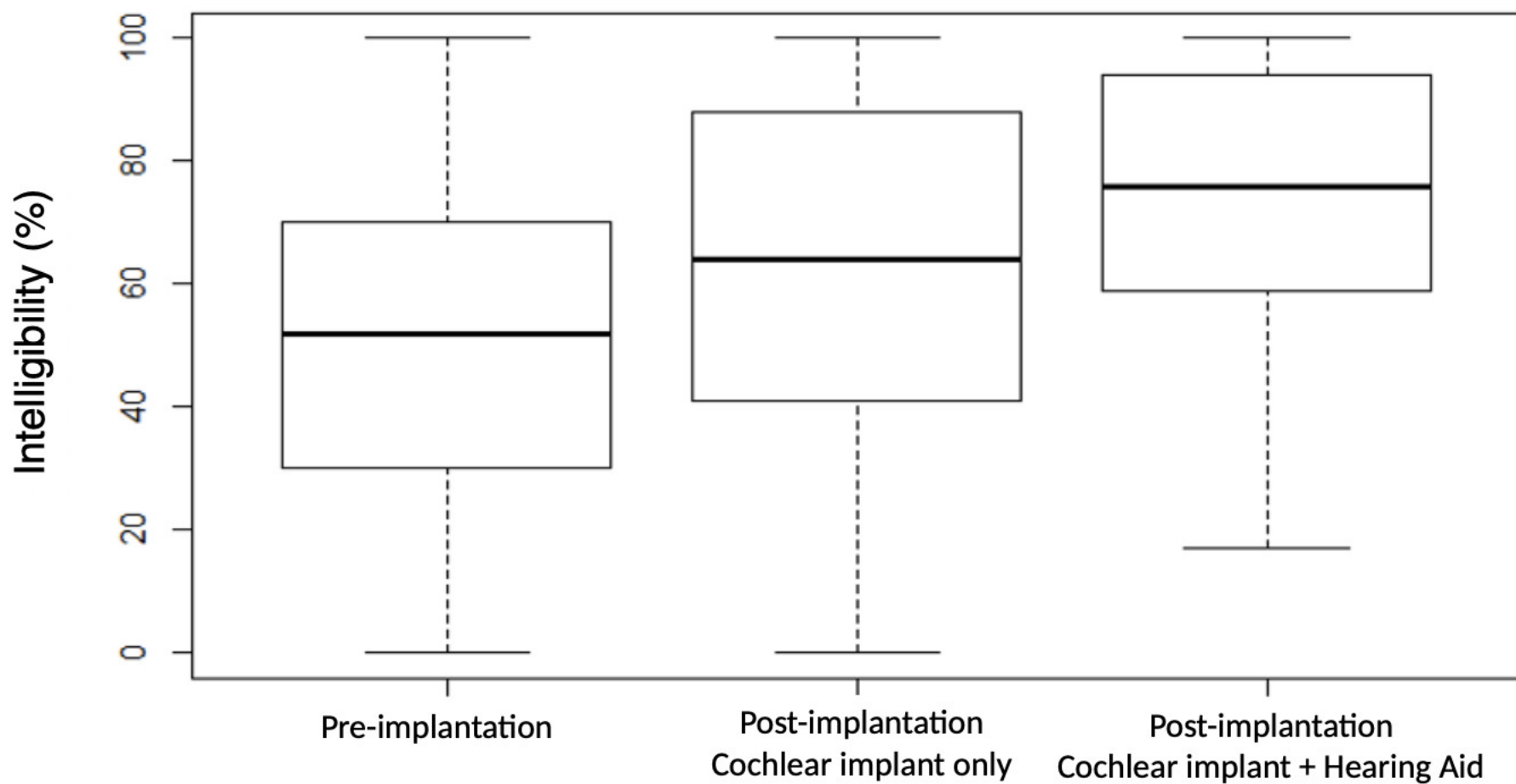
Dissyllabic word lists



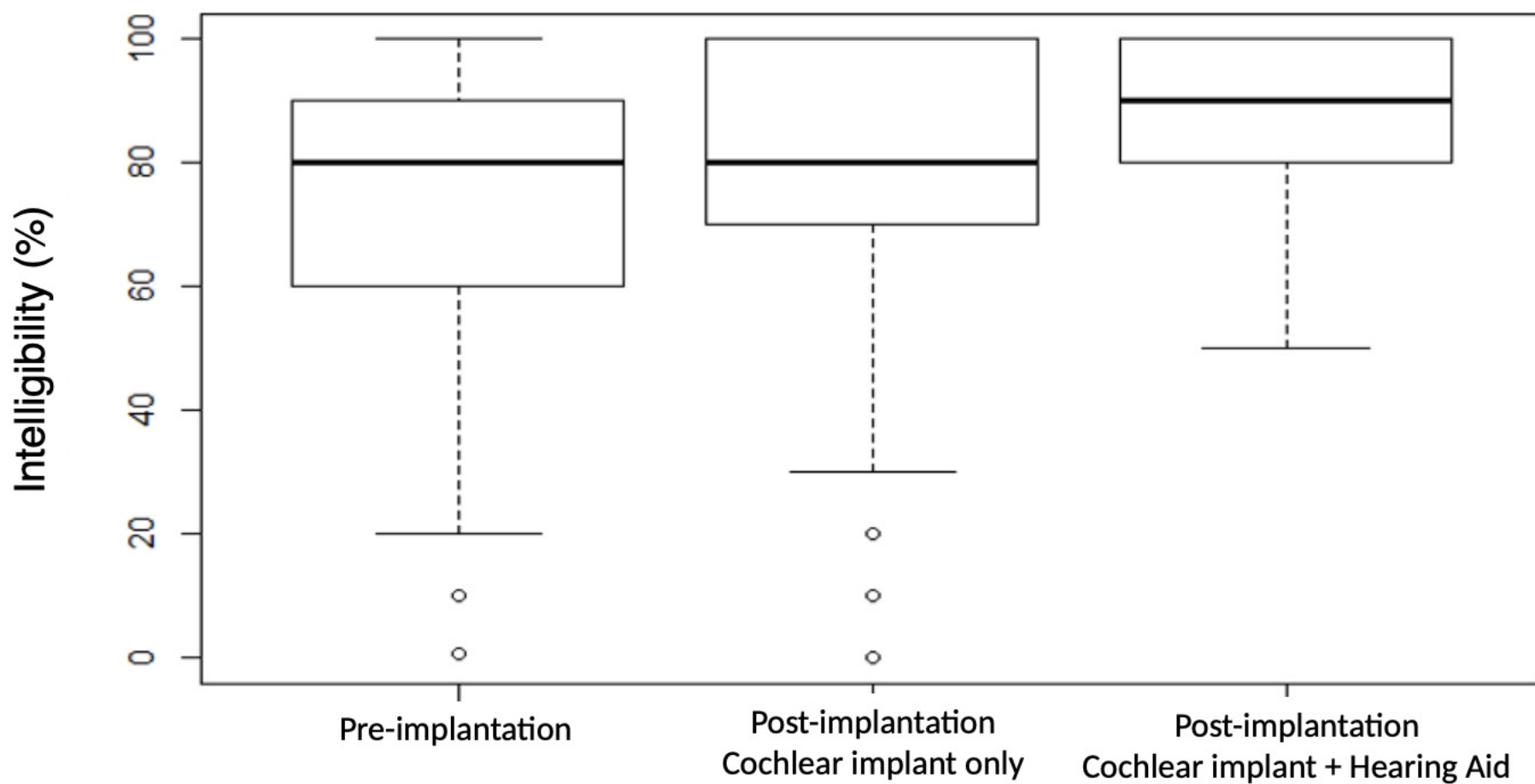




Monosyllabic word lists



Dissyllabic word lists



CAP scores

8
6
4
2

Pre-implantation

Post-implantation

