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OTOLOGY

The impact of etiology and duration of deafness on speech perception outcomes in SSD patients

Anja Kurz[1] · Marius Grubenbecher[1] · Kristen Rak[1] · Rudolf Hagen[1] · Heike Kühn[1]

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Abstract

Objective To investigate a cohort of adult single-sided deafness (SSD) patients who received a cochlear implant and to determine the impact of underlying causes of etiology and duration of deafness on outcome

Study design Retrospective data analysis

Setting Tertiary referral centre with a large cochlear implant program

Subjects and methods A demographic description of 55 adult patients implanted between 2009 and 2016. The best available speech perception score in every patient using the Freiburg Numbers, Freiburg Monosyllables and the Hochmair-Schulz-Moser (HSM) sentence test measured at the 1-, 3-, 6- and 12-month intervals, and the yearly follow-up appointments were examined. A multivariate regression analysis was conducted on the variables speech test, duration of deafness and etiology. Patients were split into four groups according to their duration of deafness (shorter duration of 10 years or less versus longer duration of more than 10 years) and etiology (inflammatory disease versus other causes).

Results The median word reception score for the Monosyllables at 65 dB SPL were 43.75% (IQR: 29.38) and 67.50% (IQR: 25.63) at 80 dB SPL at 1 year following cochlear implantation. The median percentage score correct for the HSM sentence test was 80% (IQR: 62.95). Etiology of the reviewed patient cohort revealed that most frequent causes for deafness were sudden hearing losses and inflammatory etiologies, e.g. otitis media, labyrinthitis, meningitis, cholesteatoma or mumps. The duration of deafness was not significantly associated with poor speech perception outcome. A significant correlation was found for inflammatory diseases and duration of deafness of longer than 10 years.

Conclusion The etiology and duration of deafness are important factors for the estimated outcome in speech perception in SSD patients. Presented data reveal that an inflammatory disease leading to deafness in combination with a long duration of deafness (10 + years) lead to poorer speech perception outcomes.

Keywords Single-sided deafness · Duration of deafness · Etiology

Introduction

Cochlear implantation (CI) has become a well-accepted treatment option for patients with single-sided deafness (SSD). According to Vincent et al. [1], SSD describes a hearing loss of ≥ 70 dB in the worse ear and normal hearing of ≤ 30 dB HL in the opposite ear. Asymmetric hearing loss (AHL) on the other side is defined as pure tone average in the worse ear of ≥ 70 dB HL and between > 30 dB and ≤ 55 dB hearing loss in the better ear.

The initial idea that hearing can be improved in SSD patients arose in a study in which the treatment of tinnitus in the deafened ear was the main objective [2, 3]. Since then, electrical stimulation provided via the cochlear implants has proven to facilitate localization [4, 5], hearing in noise [6] and subjective spatial awareness [7] of sounds in both SSD and AHL patients.

Today, the CI provision and rehabilitation for SSD has become a clinical standard. This leads to an increased number of adult patients arriving at our clinic with an unclear pathology and a long duration of deafness. The impact of the

Anja Kurz and Marius Grubenbecher contributed equally to this paper.

✉ Anja Kurz
Kurz_A@ukw.de

[1] Department of Oto-Rhino-Laryngology, Plastic, Aesthetic and Reconstructive Head and Neck Surgery, Comprehensive Hearing Center, University Hospital Würzburg, Josef Schneider Str. 11, 97080 Würzburg, Germany

duration of deafness has been controversially discussed in the past years. Some studies showed a negative impact on speech perception outcome with a long duration of deafness [8]; in other studies, there was no negative impact detected [9, 10]. Interestingly, the impact of the etiology of SSD has been discussed in children but only to a small extend in adults. Arndt et al. [11] reported on a trend to better speech perception outcomes in post-lingual children with a short duration of deafness compared to the group of pre-lingual implanted children. Távora-Vieira et al. [12] discussed localization abilities in post-lingual deafened adults, correlating gender, age of implantation and duration of deafness and found out that these factors did not impact localization abilities.

Despite the positive aspects of CI provision in SSD patients, one has to assume that not every patient is a suitable candidate for CI in unilateral deafness and alternatives, and, as such, the provision of a CROS hearing aid or bone-conduction device should be evaluated in all patients prior to CI. The etiology of SSD and asymmetrical hearing loss has been described by Usami et al. [13], reporting that 55% of the affected Japanese population had an idiopathic sensorineural hearing loss as an underlying cause for SSD. In 16%, the origin of disease was unknown, and, in the remaining cases, various causes, such as perilymphatic fistula, tumours, cholesteatoma otitis media or chronic otitis media, have been described. Due to the variety of underlying causes, an appropriate intervention should be considered carefully.

This retrospective data analysis aimed to investigate our SSD population receiving a CI with respect to underlying causes and outcomes. Furthermore, the goal was to identify factors which have an impact on successful speech understanding outcomes in post-lingual deafened SSD patients to improve counselling prior to CI implantation.

Materials and methods

This retrospective data analysis was performed with the approval of the local Ethics Committee of the Medical University of Würzburg.

From 2009 to 2016, 61 SSD patients were implanted in a tertiary referral centre. The established CI follow-up program comprised regular sound processor fittings at the intervals 1, 3, 6 and 12 months after initial activation, medical consultation and counselling on rehabilitation and training options.

The follow-up program in our clinic foresees also regular speech intelligibility testing at the intervals 1, 3, 6 and 12 months after the first activation, followed by a regular check-up appointment once a year. Speech perception was measured using the German Freiburg Numbers test [14] at 65 dB SPL, the Freiburg Monosyllables at 65 dB and 80 dB SPL and the Hochmair-Schulz-Moser (HSM) [15] sentence test at 65 dB SPL in quiet environments.

In all conditions, either speech was presented from the front and the contralateral ear was occluded with both a sound plug and earmuff or narrow band noise was presented via a portable audiometer or inserts.

In our clinical routine, the Freiburg numbers are tested in all hearing-impaired patients first. When the scoring is below 50% (number of items correctly understood at a sound pressure level of 65 dB SPL), no further speech testing is conducted. When the scoring comes to 100% a so-called "ceiling effect" is reached, meaning that this kind of test is too simple for the patient. Once 100% were achieved with the Freiburg numbers test, it is discontinued for the upcoming appointments; the Freiburg Monosyllables and HSM tests are then administered immediately. An irregularity in the number of tests performed may be caused by this circumstance.

Statistical analysis

Statistical analysis and a display of figures for the test results were performed using GraphPad Prism 7.0 (GraphPad Software, San Diego, CA 92,108, US) and Stata Version 14.1(StataCorp LLC, US) for regression analysis. A certified statistician (last author) supervised all statistical analyses.

Results

Patient demographics

A pre-analysis of the entire data set has shown that not all patients have passed all tests. To describe the changing number of available data sets, a general description will be presented first (Fig. 2 a–d), followed by a detailed analysis using the best-ever achieved score per patient (Figs. 3, 4.)

Of the 61 patients implanted, only 55 had documented data on speech understanding over time, and no regular documentation of speech perception outcome measurements was found in 6 patients. These 55 patients represent the total population examined and are referred to in the following as N ($N = 55$).

There were 24 (44%) male and 31 (56%) female patients. The average age at implantation was 43.65 years, with the youngest patient being 16 and the oldest 64 years of age. The inclusion of one 16-year-old patient was carried out due to the mature capacity of this individual and expected performance. In addition, most follow-up appointments occurred at and after the age of 18 years. At the time of data collection in 2016, the average age was 46.67 years (19.73–68.95 years); the average hearing experience with CI was 3.02 years (0.33–6.28 years).

The duration of deafness before implantation varied greatly; the mean value was 12.12 years with a variation between 0.08 and 55 years.

The causes for deafness and the respective frequencies in the implanted patient population ($N=61$) are shown in Fig. 1. The most common causes for deafness was sudden hearing loss ($n=21$ cases), followed by twelve patients ($n=12$) suffering from an infectious inflammatory disease, e.g. otitis media, labyrinthitis, meningitis, cholesteatoma, mumps or unspecified infection during childhood. Other causes were head trauma ($n=8$) or single incidents of deafness following surgery (e.g. saccotomy), congenital malformation, otosclerosis or progressive sensorineural hearing loss ($n=11$). In nine patients, the etiology was deemed unclear ($n=9$).

In this patient population, most patients received a CI from the manufacturer MED-EL. The implanted devices were Sonata ($n=43$), Concerto ($n=7$) and Synchrony CI ($n=5$). Five patients received the Nucleus CI512 from Cochlear; one patient was implanted with the HiResUltra implant from Advanced Bionics.

In this study, the average performance in speech perception outcomes in post-lingual deafened adults independent of their type of implant and sound processor was assessed.

To filter out the influence of the multiple measurements on the respective examination periods, the best measurement result obtained per time unit and the patient was used for the analysis in the following figures.

Speech audiometry

Freiburg numbers

The Freiburg number test at 65 dB was carried out in 53 patients ($N_{total}=53$). Eighty percent of the measurements were performed in the first year after implantation. Figure 2a denotes the median of the best results in the Freiburg number test for the respective investigation periods. When measured after both the first 3 and 6 months, the median was at 75% (IQR: 72.5 and 74.37, respectively) and, after 1 year, at 90% (IQR: 22.5). In the second year after implantation, the median was at 100% (IQR: 70) and 75% after the third year (IQR: 25). After 4 years, the median sloped upwards to 92.5% (IQR: 56.25).

Freiburg monosyllables

The Freiburg Monosyllables at 65 dB were accomplished in 49 patients ($N_{total}=49$). Figure 2b shows the median of the best results in the respective examination periods. The median was 25% (IQR: 31.25) for up to 3 months after implantation and increased to 36.25% (IQR: 37.5) for measurements taken between 3 months and half a year. In the next investigation period (up to 12 months after implantation), the median increased further to 43.75% (IQR: 29.38), and, in the period up to 2 years after implantation, the median dropped to 30% (IQR: 33.75). Between the second and third years after implantation, the median decreased again to 25% (IQR: 55.26). In the fourth year after implantation, the median mounted to 43.75% (IQR: 50) and decreased to 28.75% (IQR: 60.63) in the fifth and sixth years after implantation.

The Freiburg Monosyllables at 80 dB was conducted in 47 patients ($N_{total}=47$). Most test results were available for 18 patients in the second year after implantation.

Figure 2c shows the median of the best results for Monosyllables (80 dB SPL) for the respective examination periods.

The median in the first measurement period (up to 3 months) was 43.75% (IQR: 31.25). In the measurement period of up to half a year, the curve sloped upwards to 50% (IQR: 53.13). At the 1-year interval, the median was 67.5% (IQR: 25.63); in the second year, 62.5% (IQR: 28.75) and, in the third year, 62.5% (IQR: 37.5). In the fourth year, the curve decreased to 40% (IQR: 73.75). Lastly, for the fifth and sixth years together, the curve increased again to a median of 61.25% (IQR: 58.12).

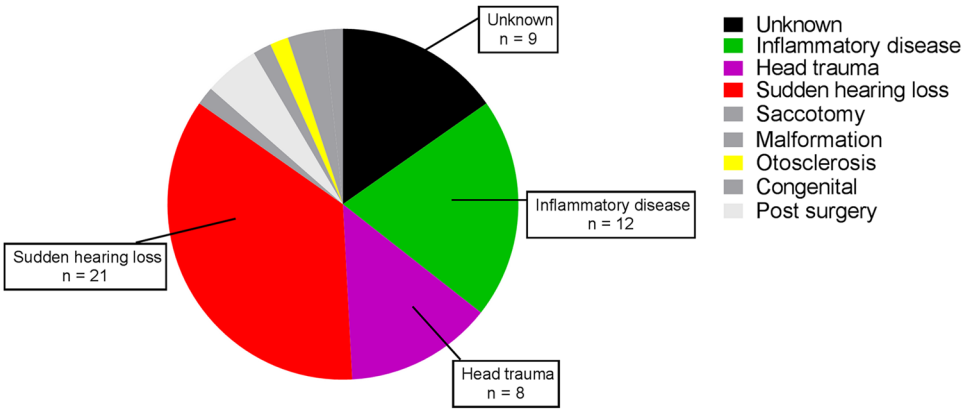

Fig. 1 The causes of deafness in the adult SSD population (2009–2016)

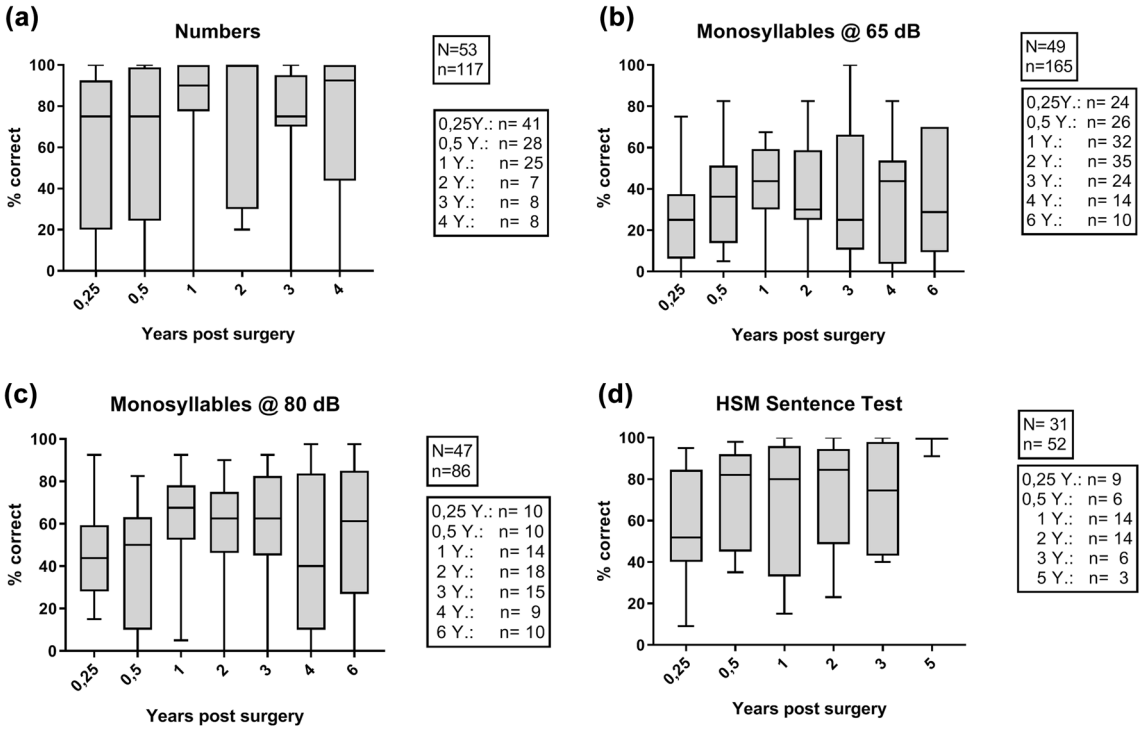

Fig. 2 The boxplots (min to max) denote the medians in percent correct (%) for the **a** Freiburg numbers test **b** Freiburg monosyllables at 65 dB SPL **c** Freiburg monosyllables at 80 dB SPL and **d** HSM-sentence test over a period of 4–6 years. N represent the total amount of patients, n the available data sets

HSM sentence test

The HSM sentence test was accomplished in 31 patients ($N_{total} = 31$) and, overall, was used less frequently than the Freiburg tests.

Figure 2d shows the median of the results in the HSM sentence test for the respective examination periods. The medians went up in the first half-year after implantation and then reached a plateau. For the period of up to 3 months after implantation, the median was at 51.85% (IQR: 44.5). For measurements between 3 months and half a year, the median increased to 82% (IQR: 47), and between 6 months and 12 months, the median was at 80% (IQR: 62.95). In the second year after implantation, the median was at 84.5% (IQR: 45.95); in the third year at 74.5% (IQR: 54.9) and, in the fourth and fifth years, at 100% (IQR: 9).

The impact of hearing experience on speech perception outcome

A detailed analysis was further conducted in the upcoming two figures using the best-ever achieved score per patient. To provide a more detailed view on the impact of hearing experience, the best-achieved speech perception score per patient was determined and plotted against a time scale.

Figure 3a illustrates that most patients were capable of scoring between 60–100% within the first 12 months after implantation. This trend is less visible when investigating the Monosyllabic words (see Fig. 3b, c).To score more than 50% correct for Monosyllables, clients needed, on average, 24 months (at 65 dB SPL) and up to 72 months for louder speech items (80 dB SPL). Results of the HSM sentence test (Fig. 3d) showed that successful speech perception varies greatly but could be achieved in some cases even after 36 months.

The impact of duration of deafness and etiology

The impact of duration of deafness in our patient collective provides another source of variability. In the analysed population, the duration of deafness ranges between less than 1 year and 55 years. The mean value is 12 years, the median is 4 years and the interquartile interval is 19. Forty percent of the patients have a duration of deafness of less than 2 years. When classified into less and more than 10 years of deafness, groups of 35 (64%) and 20 (36%) patients, respectively, were established.

Of the 55 patients in the total population, six were prelingual deafened, which means they deaf before the age of two. Due to their early deafness, these patients are part of the group of patients with a long duration of deafness.

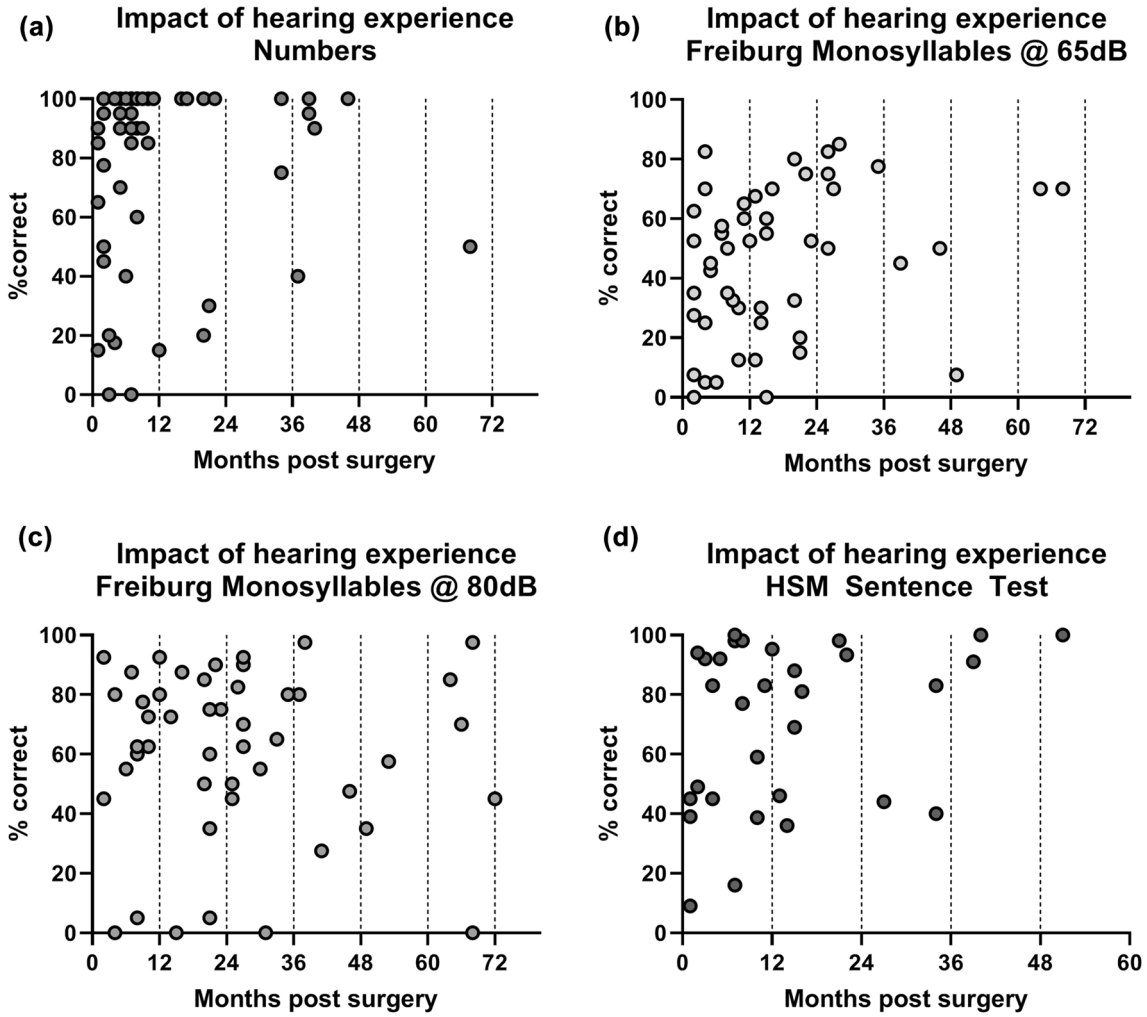

Fig. 3 Impact of hearing experience on speech perception for **a** Freiburg numbers, **b** Freiburg monosyllables at 65 dB, **c** Freiburg monosyllables at 80 dB and **d** HSM sentence test. Single points denote the time needed to achieve the best-documented individual speech score

Since the main reason of the analysis was the question regarding the duration of deafness and not the issue of whether pre-lingually deaf ears can be rehabilitated, the group of pre-lingual deaf patients was excluded in the further analysis.

Figure 4a, b depict the effect of deafness on hearing performance after CI. When examining the variable "deafness duration", it became apparent that within the group with a deafness duration of more than 10 years, the values were not distributed around the median, but, rather, two groups were emerging. Patients with inflammatory etiology performed worse. The following etiologies were categorized under inflammatory disease: otitis media, labyrinthitis, meningitis, cholesteatoma, mumps and unspecified infection during childhood.

Accordingly, in Fig. 4a, b a distinction is made between four groups:

A = inflammatory disease and less than 10 years duration of deafness.

B = inflammatory disease and 10 or more years of exposure to deafness.

C = other cause and less than 10 years of deafness.

D = other cause and ten or more than 10 years of deafness.

Evaluating the Freiburg Monosyllables according to the above-mentioned classification, the number of patients per defined group differs. Of the 49 patients in total, there are 3 patients in Group A (6%), 7 patients in Group B (14%), 31 patients in Group C (63%) and 8 patients in Group D (16%). The median is 82.5% in Group A (IQR = 47.5), 12.5% in Group B (IQR = 15), 52.5% in Group C (IQR = 40) and 53.75% in Group D (IQR = 22.5). The significantly lower median in Group B in contrast to the other groups is particularly noticeable here. The mean value provides similar results to the median and is 78.33 in Group A (SD = 24.02), 11.79 in Group B (SD = 8.981), 49.44 in Group C (SD = 24.09) and 54.69 in Group D (SD = 13.19). The Kruskal–Wallis test was performed for statistical analysis to find a significant difference between

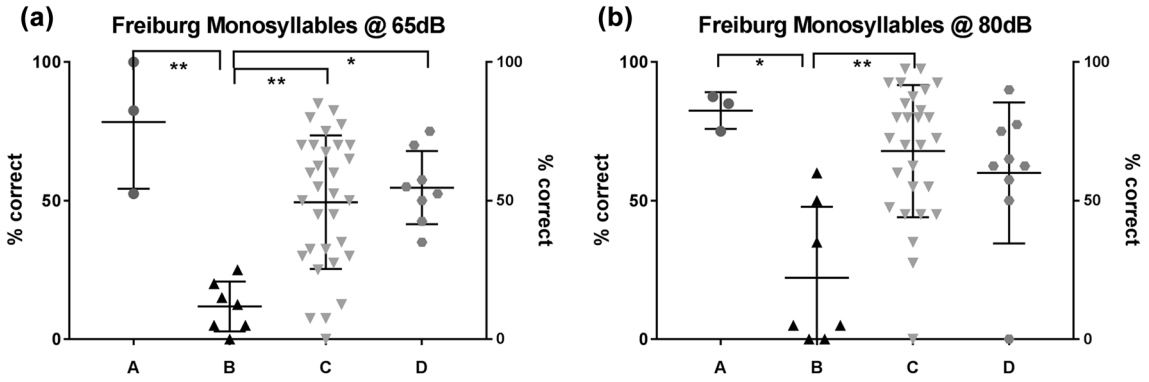

Fig. 4 Effect of the duration of deafness (± 10 years) and etiology (inflammatory disease vs. other causes) on speech perception outcome (% correct) using linear regression analysis. Group A = inflammatory disease ≤ 10 years of deafness; Group B = inflammatory disease ≥ 10 years of deafness; Group C = other cause of deafness ≤ 10 years deafness duration; Group D = other cause of deafness ≥ 10 years deafness duration. *$p < 0.05$; **$p < 0.01$

the groups (X^2 ($N = 49$) = 16.27; $p = 0.0010$). Dunn's comparisons test was carried out as a post-hoc test. Group B differed significantly from all other groups. The corrected p-value (due to multiple measurements) between Groups A and B was $p = 0.0031$, $p = 0.0050$ between B and C and $p = 0.0153$ between B and D.

The results with the Freiburg monosyllables at 80 dB are also distributed differently among the groups. A total of 47 values ($N = 47$) was redistributed to Group A with 3 patients (6%), Group B 7 patients (15%), Group C 28 patients (60%) and Group D 9 patients (19%). In Group A, the median was 85% (IQR = 12.5), in Group B 5% (IQR = 50), in Group C 72.5% (IQR = 37.5) and in Group D 62.5% (IQR = 22.5). The mean value was 82.5% in Group A (SD = 6.614), 22.14% in Group B (SD = 25.63), 67.86% in Group C (SD = 23.82) and 60% in Group D (SD = 25.43). Again, the number of data in Groups A and B were too low for the D´Agostino & Pearson test, so no normal distribution was assumed. The Kruskal–Wallis test determined a significant difference between the groups (X^2 ($N = 47$) = 12.73; $p = 0.0053$). The Dunn´s comparisons test showed a significant difference between Group B and Groups A and C. The corrected p-value between Groups A and B was $p = 0.0244$ and $p = 0.0073$ between B and C.

Multivariable linear regression analysis

To test the influence of different factors on the outcome of speech testing in a statistical test procedure at the same time, a multiple linear regression for each speech test was conducted (see Table 1). The dependent variable is the result of the respective speech test (Freiburg Number, Monosyllables, HSM test). The selected independent variables are hearing experience, duration of deafness and etiology of hearing loss (inflammatory disease).

Table 1 shows, for the Freiburg numbers, a p-value of 0.046 for the variable inflammatory disease without considering the duration of deafness. Combining the variable inflammatory disease and deafness duration, there is a significant negative influence ($p = 0.012$) on the outcome in the Freiburg number test.

Another statistically significant effect was detected also in the Freiburg Monosyllables at 65 dB; here, the p value is again significant for the variable inflammatory disease ($p = 0.018$) and highly significant ($p = 0.000$) for the combination inflammatory disease and deafness duration.

For the Freiburg Monosyllables at 80 dB, both hearing experience ($p = 0.019$) and inflammatory disease ($p = 0.005$) as significant variables. The strongest correlation is again for inflammatory disease and duration of deafness ($p = 0.000$).

In the HSM test, significant correlations are found for hearing experience ($p = 0.001$), duration of deafness ($p = 0.047$) and the combination inflammatory disease and duration of deafness ($p = 0.066$).

Discussion/Conclusion

In the past years, we encountered a number of SSD patients who had only limited benefits with a CI; therefore, it was necessary to look for the underlying reasons. One of the suspected underlying causes was long-term deafness, and the other suspicion was no or too few auditory training efforts.

To begin, the factor duration of deafness has been controversially discussed in literature. There are large statistical studies, e.g. Holden et al. [16], in which the duration of deafness was identified to be negatively correlated with speech

Table 1 Multivariable linear regression analysis

	Multivariante linear regression analysis			
	Variable	β	p	SE
Freiburg numbers	Hearing experience	0.332	0.059	0.172
	Inflammatory disease	12.964	0.046 *	6.354
	Duration of deafness	− 19.158	0.133	12.567
	Inflammatory D. and duration of deafness	− 38.859	0.012*	14.869
Freiburg monosyllables @ 65 dB SPL	Hearing experience	0.159	0.153	0.109
	Inflammatory disease	26.360	0.018*	10.776
	Duration of deafness	− 0.283	0.955	5.033
	Inflammatory D. and duration of deafness	− 55.059	0.000**	11.795
Freiburg monosyllables @ 80 dB SPL	Hearing experience	0.365	0.019*	0.150
	Inflammatory disease	21.627	0.005*	7.366
	Duration of deafness	− 1.677	0.814	7.076
	Inflammatory D. and duration of deafness	− 60.29	0.000**	12.196
HSM test	Hearing experience	0.677	0.001*	0.183
	Inflammatory disease	7.398	0.606	14.196
	Duration of deafness	− 17.998	0.047*	8.683
	Inflammatory D. and duration of deafness	− 32.555	0.066*	17.050

B Pearson standardized regression coefficient, *SE*, standard error, *p* significance (*$p < 0.05$, **$p < 0.001$)

perception outcome in bilaterally deaf CI patients. Other authors, e.g. Moon et al. [17], described that the age at the onset of deafness was closely related to the postoperative performance but argue that duration of deafness should not be the sole indicator for exclusion from CI surgery, in general. Medina et al. [18] supported the claim that long-term auditory deprivation in the ear to be implanted should not be the criterion to reject CI.

These studies are not consistent with our daily clinical experience.

In literature, the indication group "SSD" has undergone controversial discussions on the topic duration of deafness. In a recent publication by Cohen and Svirsky [19], the authors conclude that speech perception scores in SSD patients are negatively correlated with the duration of deafness. However, there is still uncertainty due to the limited amount of strong statistical data which precludes clinical recommendations. Cohen and Svirsky performed a linear regression analysis in eight selected publications and excluded visual outliers in the available speech tests. By excluding the outliers, three previously significant correlations lost their significance, which indicated that there is still limited statistical power in the published studies. Cohen and Svirsky further discussed if a period of 5 or 10 years of auditory deprivation may predict poorer speech perception. In our patient population, we found a large variability concerning the duration of deafness (ranging from below 1 year to 56 years). When starting with a first statistical analysis we did not see effects when establishing patients in duration-of-deafness groups of 0–5 years and 5–10 years. The strongest effects could be seen when determining a 10-year auditory deprivation period and greater.

Even today, there are SSD patients who present to our clinic with an unclear etiology and the question if the onset of hearing loss war pre-, peri- or post-lingual. In our cohort, we excluded the known congenital patients to establish the single-factor duration of deafness and underlying causes.

Analysis of the actual results supports the idea that duration of deafness is not the only indicator for a prediction of outcome with CI in SSD patients. In addition, inflammatory disease as the underlying cause in combination with a long duration of deafness (more than 10 years) is a negative predictor. Interestingly, the three patients allocated to Group A (inflammatory disease and short duration of deafness) showed an overall good performance in speech understanding. Considering Group C, the combination of "other causes of deafness" and "short duration of deafness", revealed a high variance in outcomes. A long duration of deafness alone does not imply an overall poor speech understanding, as shown in Group D.

On the contrary, present data show that a long duration of deafness and an inflammatory disease (Group B) have a negative influence on speech perception outcomes.

The impact of meningitis or mumps on CI outcome has been addressed in the scientific medical community. It is known that meningitis leads to cochlear ossification; hence, an early CI implantation following the disease is recommended. Helmstaedter et al. [20] evaluated speech perception outcomes in 35 CI-implanted children who lost their

hearing due to bacterial meningitis. Their study investigated, furthermore, the impact of electrode and implant charge in unaltered, obliterated and ossified cochleae. It was demonstrated that an increased charge was needed for higher cochlear implant output. A reduced audiological outcome was found in the group with obliterated and ossified cochlea. The investigated patient-cohort in our retrospective data analysis included two post-meningitis patients and two patients who became deaf due to a mumps infection (during childhood); they had no ossification or other pathologies, as verified by pre-operative CT/MRT scans.

The impact of underlying diseases on CI outcome has further been discussed by Katsushika et al. [21]. The authors investigated the impact of mumps on speech perception in four pediatric cases, concluding that early CI resulted in good speech perception whereas late intervention led to a poorer outcome. A case study with three patients by Hassepass et al. [22] reported on speech perception outcomes after labyrinthitis ossificans and concluded that early intervention with a CI increased the likelihood for post-implant success. In our patient cohort, of the twelve patients that were unilaterally deafened due to an inflammatory disease, only three became good performer and their duration of deafness was below 10 years. Based on our findings, we emphasize an immediate cochlear implant intervention in deaf patients with an inflammatory etiology to provide the chance for successful rehabilitation with CI. There is the need to confirm our findings in larger studies and to underpin the impact of etiology and duration of deafness.

Despite the underlying cause and duration of deafness, the conducted multivariate linear regression analysis calculated the factor "hearing experience" to be significant, as well (Monosyllables at 80 dB SPL, HSM sentence test). Figures 2 a–d depict that, on average, the best results were achieved within the first 2 years after implantation. However, there are patients who reach their optimal performance after 60–72 months. It can be anticipated that this late success is only possible due to regular auditory training. In our CI program, counselling on auditory training has been implemented pre-operatively and post-operatively at several occasions (first activation and 1-,3-,6-month post-op). During these counselling appointments, we reinforce the use of the direct audio input to train the CI-implanted ear following a structured auditory training program. Additionally, another integral part of the program is to set realistic expectations; therefore, we emphasize the importance of regular auditory training over the upcoming years in all patients.

Based on the present findings, it can be concluded that the underlying etiology and the duration of deafness are important predictors for speech outcome in SSD patients. Patients with a long duration of deafness and an inflammatory cause of deafness should be advised to try other hearing devices, such as CROS or bone-conduction hearing aids, as an alternative. Patients deafened in the course of an inflammatory disease are advised to proceed shortly after diagnosis with the clinical pre-CI testing procedure to facilitate early CI implantation.

Compliance with ethical standards

Conflict of interest None of the authors received funding in relation to this article. The authors declare that they have no conflict of interest.

Ethical approval All procedures performed in this study involving human participants were in accordance with ethical standards of the institutional and /or national research committee and with the 1964 Helsinki declaration. Because of the retrospective study design, formal consent was not required. The study was approved by the Ethical Commission of the Medical University of Würzburg, Germany (Nr. 20180808 02).

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