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ORIGINAL RESEARCH

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Performance and self-perceived hearing impairment after cochlear implantation in Menière's disease

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Abstract

Objective: Evaluation of the self-perceived hearing impairment and performance after cochlear implantation in patients with definite Menière's disease (MD).

Patients and Methods: Seventeen unilaterally or bilaterally profoundly hearing-impaired patients suffering from MD who received a cochlear implantat (CI) were eligible for inclusion in this study. Their self-perceived hearing impairment using the short *Speech Spatial and Qualities of Hearing Scale* (SSQ12) as well as their performance in speech perception (German language Freiburger mono- and multisyllable test, Oldenburger sentence test) were compared with a best-matched control group of non-MD patients up to 24 months of follow-up.

Results: MD patients improved significantly in perception of monosyllables presented at 65 dB_{SPL}, from preoperatively best aided 18.2% [2.4, 34.0] to 51.7% [39.4, 63.9] 1 year after cochlear implantation (mean [95% confidence interval]). Their performance approached the matched controls with 63.2% [55.7, 70.8]. Monosyllables presented at a lower intensity of 55 dB_{SPL} revealed a significant underperformance of the MD patients (21.1% [12.6, 29.6]) in contrast to the non-MD controls (39.1% [30.9, 47.4]) 12 months post-CI. Self-assessed hearing disability was significantly more pronounced in MD patients with a mean total SSQ12 score of 3.6 [2.4, 4.9] in comparison to 6.1 [5.4, 6.8] of the matched non-MD controls after 12 months of cochlear implantation.

Conclusion: Cochlear implantation substantially improves hearing capabilities in profoundly hearing-impaired patients with MD, but they tend to underperform in comparison to non-MD patients at least at lower sound pressure levels. This is likely one reason for the poorer self-assessed hearing function of cochlear implanted MD patients.

Level of Evidence: 3, retrospective, nonrandomized follow-up study.

KEYWORDS

cochlear implant, deafness, hearing impairment, hearing loss, hearing outcome, Menière's disease, speech recognition, SSQ12

1 | INTRODUCTION

Menière's disease (MD) is a multifactorial disorder of the inner ear comprising the symptoms vertigo, tinnitus, sensorineural hearing impairment, and aural fullness as an episodic progressing clinical syndrome.[1] MD is strongly associated with an endolymphatic hydrops (increased pressure in the membranous labyrinth) but the definitive pathophysiology still remains unclear.[2]

The clinical course of MD, as well as the manifestation of the different symptoms, show considerable variation among patients. From the onset of the disease, a fluctuating sensorineural hearing loss (SNHL) progresses, especially in the low frequencies, and usually stabilizes to moderate-to-severe hearing impairment within 5–10 years.[3] In a substantial proportion of patients, however, SNHL progresses to a profound stage at which cochlear implantation is a viable treatment for hearing restoration (recently reviewed in References 4 and 5).

There are currently few data in the literature on self-perceived hearing function of MD patients after cochlear implantation. A validated tool to evaluate self-assessed hearing impairment, also in implanted patients, is the SSQ12,[6,7] a more practical short form of the original *Speech Spatial Qualities of Hearing Scale* (SSQ49).[8] It is one of the most frequently applied self-reported measures of hearing function, and is composed of several listening capability items of three different categories.

The current retrospective study of cochlear implanted patients suffering from unilateral or bilateral MD examined self-perceived hearing impairment by the use of the SSQ12 in relation to word- and sentence testing with up to 24 months of follow-up, in comparison to a matched, cochlear-implanted control group.

2 | PATIENTS AND METHODS

2.1 | Study

This study used a retrospective case–control design, and approval was obtained from the local Ethics Board of the University of Göttingen (nr. 24/2/21). Differences in self-reported hearing impairment (SSQ12) between MD patients and matched controls were the primary outcome, differences in speech perception (word and sentence tests) the secondary outcome. General personal and disease-related data were collected from the review of patient records, specific audiological data from preoperative testing, first fitting and from follow-up examinations and adjustments, which were regularly performed 3, 6, 12, and 24 months after the initial cochlear implant (CI) adjustment.

2.2 | Patients

All patients who received a cochlear implant at the Department of Otorhinolaryngology of the University Medical Center Göttingen between 2009 and 2019 were screened for a possible diagnosis of MD. Subsequently, patient's charts were reviewed to determine whether they met the criteria of definite MD according to the current International Classification of Vestibular Disorders diagnostic criteria.[1] Additionally, existing imaging data (magnet resonance imaging as well as computer tomography of the head) was reevaluated to further exclude underlying pathologies other than MD.

For every MD patient, the best two available control matches regarding age at implantation as well as duration and progression of hearing loss as well as hearing thresholds were identified to provide a control collective of non-MD patients. Controls were recruited from the same collective, implanted between 2009 and 2019. Preoperative vertigo, as well as documented vestibular dysfunction before implantation, were exclusion criteria.

Patients of the MD and non-MD group were supplied with cochlear implants manufactured by MED-EL, Cochlear as well as Advanced Bionics, detailed information regarding the specific implants can be obtained from Table S1 (supplementary material).

2.3 | Subjective questionnaires

The short form (SSQ12) of the *Speech Spatial Qualities of Hearing Scale* was employed to obtain the self-assessment of hearing disability subdivided into speech perception, spatial-, and quality auditory perception. The original SSQ is a validated instrument to measure hearing function (also in cochlear implanted patients) and results of the short form SSQ12 are strongly correlated.[6–10] Advantages of the SSQ12 are that the short form is less time consuming, better accepted by patients, and therefore more practical in the clinical routine.

2.4 | Audiologic testing

Audiologic testing was performed in a double-walled, sound-attenuating booth (DIN ISO 8254). The AT1000 (AURITEC GmbH, Hamburg Germany; EN 60645) served as audiometric test device for pure-tone audiometry (EN ISO 8253), as well as word- and sentence testing (EN ISO 8253-2) as described below.

The German language Freiburger word recognition test for multisyllables and monosyllables was performed prior to cochlear implantation with and without best-aided hearing devices (65 dB and above), as well as in every follow-up in a CI-only setting at 55 and 65 dB sound pressure level (SPL) in a quiet sound field. In follow-up examinations, sentence recognition was tested by the Oldenburger sentence test (OLSA) in quiet and noise (signal and noise from the front, 0° azimuth). The ear contralateral to testing was attenuated by using foam earplugs (SNR = 36 dB, EN352-2:2002) and/or acoustic earmuffs (3M Peletor X3A, SNR = 33 dB, EN397:1995), depending on the degree of asymmetry.

2.5 | Statistical analysis

The open-source statistics software "R" (packages: Hmisc, ggplot2, ggpubr, reshape2, dplyr) was employed for statistical and graphical

data analysis. Data were tested for normal distribution by performing the Shapiro–Wilk test. Differences between the study groups were determined by utilizing a *t* test (two-sided, unpaired) for normally distributed data, and a Wilcoxon rank-sum test (two-sided, unpaired) for non-normally distributed data. The alpha level was set to .05 (*).

3 | RESULTS

3.1 | Patient data

Of all CI-recipients between 2009 and 2019 at our center, 17 patients were detected suffering from MD and who met the inclusion criteria for enrollment in this study. Baseline and disease-specific characteristics of both study groups are listed in Table 1. The mean time between onset of MD and CI surgery was 14.2 years (±11.5 SD) and did not differ significantly ($p = .47$) from the duration of diseases of the controls (16.3 years [±11.0 SD]).

Prior to cochlear implantation, three MD patients underwent intratympanic application of gentamicin, five patients intratympanic application of lidocaine. In one MD patient, a partial labyrinthectomy was performed simultaneously to the CI surgery. Out of the 17 patients suffering from MD, 13 patients were treated with betahistine; in the other four patients, this therapy was stopped due to associated side effects. More detailed demographic and clinical information of individual patients can be obtained from Table S1 (supplementary material).

3.2 | Audiometry

The averaged preoperative pure-tone audiogram and the appropriate 95% confidence interval of all MD patients and of the control group are depicted in Figure 1. Significant differences were found at 2 ($p = .007$), 6 ($p = .049$), and 8 kHz ($p = .002$), with 81.3 dB_{HL} [67.2, 95.4] versus 102.4 dB_{HL} [92.9, 111.9], 89.6 dB_{HL} [74.7, 104.5] as compared to 105.8 dB_{HL} [95.4, 116.1] and 93.7 dB_{HL} [78.4, 109] versus 119.8 dB_{HL} [111.9, 127.7], respectively. Aided threshold measures in an open sound field 1 year after cochlear implantation revealed an almost congruent performance of the implanted side of both groups (Figure 1).

Patients suffering from MD showed a significant improvement ($p = .012$) in the perception of monosyllables presented at 65 dB_{SPL}, from 18.2% [2.4, 34.0] preoperatively best aided, compared with 51.7% [39.4, 63.9] 1 year after cochlear implantation. The control group improved in monosyllable perception (65 dB_{SPL}, $p < .001$) as

TABLE 1 Baseline characteristics

	Menière group	Control group	*p*-Value
Group	$n = 17$	$n = 34$	
Sex			.55
Male	$n = 7$	$n = 17$	
Female	$n = 10$	$n = 17$	
Mean age (years ± SD) at			
Implantation	67.8 (±7.9)	66.9 (±7.4)	.74
CI indication	64.7 (±8.2)	65.0 (±8.6)	.91
Disease begin[a]	53.5 (±13.5)	50.6 (±12.7)	.48
Diagnosis			
Bilateral MD	$n = 11$		
Unilateral MD	$n = 6$		
Sudden unilateral SNHL		$n = 8$	
Sudden bilateral SNHL		$n = 2$	
Sudden recurrent unilateral SNHL		$n = 1$	
Sudden recurrent bilateral SNHL		$n = 3$	
Progressive unilateral SNHL		$n = 1$	
Progressive bilateral SNHL		$n = 19$	
Side of CI			.24
Right	$n = 10$	$n = 12$	
Left	$n = 5$	$n = 18$	
Both	$n = 2$	$n = 4$	
Daily use of CI (hours ± SD)	12.9 (±3.3)	13.5 (±2.1)	.92

[a]Disease begin was defined as the first onset of hearing loss on the affected side.
Abbreviations: CI, cochlear implant; MD, Menière's disease; SNHL, sensorineural hearing loss; SD, standard deviation.

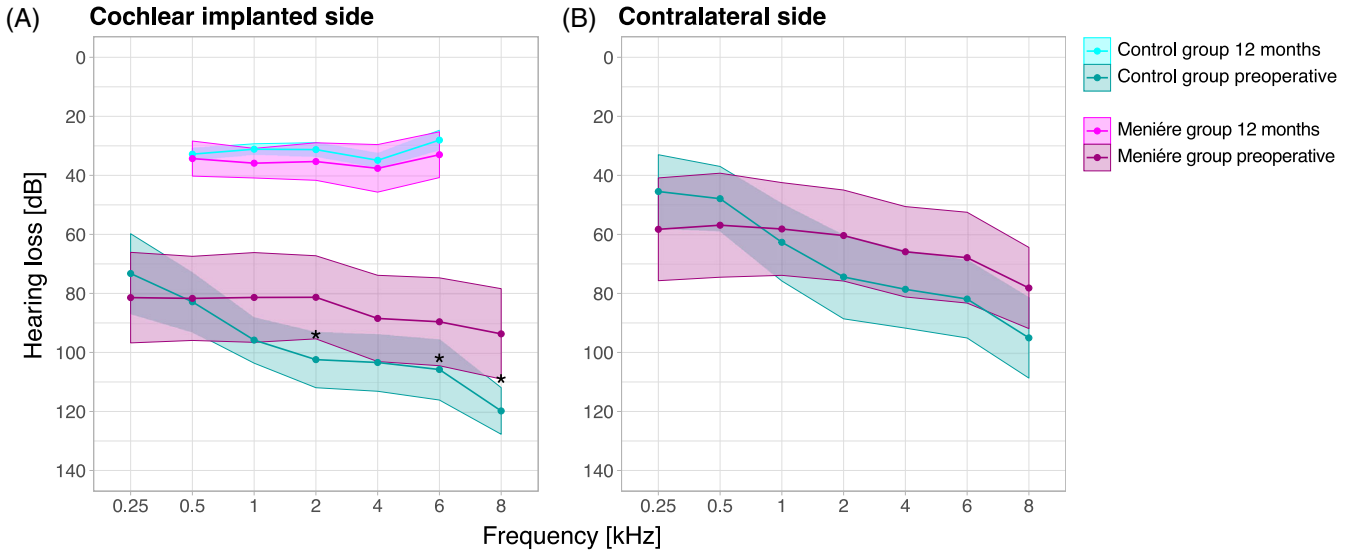

FIGURE 1 Pure-tone audiometry. Cumulative results of the preoperative pure-tone audiometry of the Menière (purple) and control group (turquoise) depicted separately for the cochlear implanted (A) and the contralateral side (B). Additionally, the cumulative pure-tone audiometric results of both groups 12 months after cochlear implantation are displayed in A. Results are given in means (points and line) as well as 95% confidence intervals (light colored area around the mean). An asterisk (*, $p < .05$) indicates significantly different hearing thresholds between groups for specific frequencies

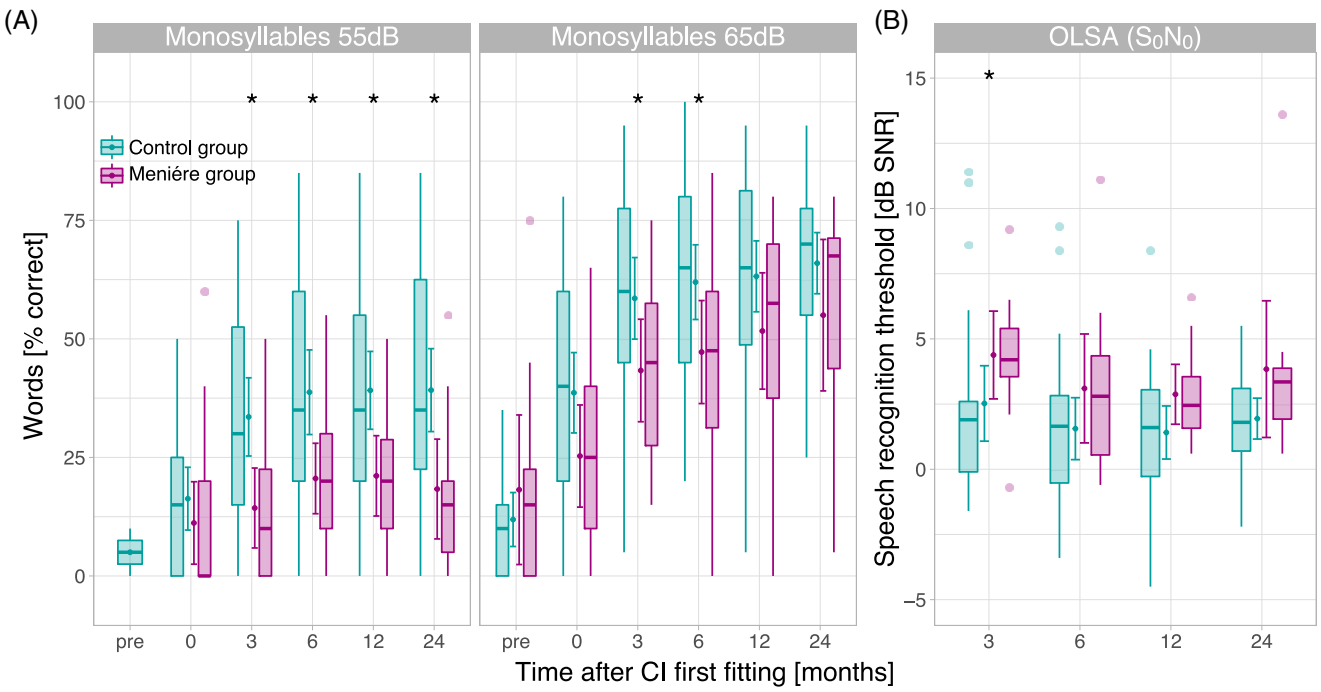

FIGURE 2 Word and sentence testing. (A) Cumulative results of the preoperative and postoperative follow-up monosyllable understanding (Freiburger word test) presented at 55 dB_{SPL} (left) and at 65 dB_{SPL} (right) of the Menière (purple) and control group (turquoise). (B) Cumulative results of the postoperative follow-up sentence testing in noise (Oldenburger sentence test), of both groups. Results are presented as the means (points) and 95% confidence intervals (error bars) as well as a standard box plot. Asterisks (*, $p < .05$) indicate significant differences between groups for specific points in time (CI follow-up)

well, from 11.9% [6.2, 17.6] preoperative to 63.2% [55.7, 70.8] 1 year after implantation. Preoperative presentation of monosyllables at 55 dB_{SPL} was not performed regularly.

Figure 2(A) shows that MD patients performed significantly worse in understanding soft monosyllables presented at 55 dB_{SPL} at every time point after cochlear implantation, except 1 month after implantation

immediately following the initial CI adjustment (3 months: 14.3% [5.9, 22.8] vs. 33.5% [25.3, 41.8], $p = .005$; 6 months: 20.6% [13.1, 28.0] vs. 38.8% [29.8, 47.7], $p = .012$; 12 months: 21.1% [12.6, 29.6] vs. 39.1% [30.9, 47.4], $p = .009$; 24 months: 18.3% [7.8, 28.9] vs. 39.2% [30.5, 47.9], $p = .008$). In contrast, at the standard speech audiometry test intensity of 65 dB_{SPL} MD patients improved their understanding of monosyllables and caught up with the control group within the first year after implantation. They thus showed no significant differences at 12 months after cochlear implantation (51.7% [39.4, 63.9] vs. 63.2% [55.7, 70.8], $p = .12$) as well as after 24 months (55.0% [39.0, 71.0] vs. 66.0% [59.5, 72.4], $p = .25$).

A similar tendency of convergence over time between MD patients and the control group was seen in the results of the sentence recognition tests in noise (OLSA, Figure 2(B)). A significant gap ($p = .03$) between the two groups was detected 3 months after cochlear implantation, being 4.4 dB [2.7, 6.1] and 2.5 dB [1.1, 3.9] in the MD group and the control group, respectively. One year after cochlear implantation, the test results of MD patients in sentence recognition improved to an average of 2.9 dB [1.7, 4.1], which was insignificantly different ($p = .11$) from the controls (1.4 dB [0.4, 2.4]).

Within the MD group, no significant differences in word and sentence testing could be found in patients who reported vertigo in the first year after cochlear implantation in comparison to patients without complaints (see supplement Figure S1).

3.3 | Self-perceived hearing function

Subjective hearing function was assessed employing the short *Speech Spatial and Qualities of Hearing Scale (SSQ12)*. The subjective hearing function of both groups improved considerably 3 months after implantation of a CI and remained relatively constant during the observation period of 24 months (Figure 3). The baseline SSQ12 mean score, captured at the beginning of the initial CI fitting, amounted to 1.64 [0.8, 2.5] and 2.1 [1.8, 2.4] for MD patients and the control subjects, respectively, and increased significantly to 3.6 [2.4, 4.8] and 6.1 [5.4, 6.8] after 12 months of implantation ($p = .011$ and $p < .001$, respectively). Figure 3(A) depicts the average scores of single SSQ12 items over time in a heatmap, separated for MD and control patients

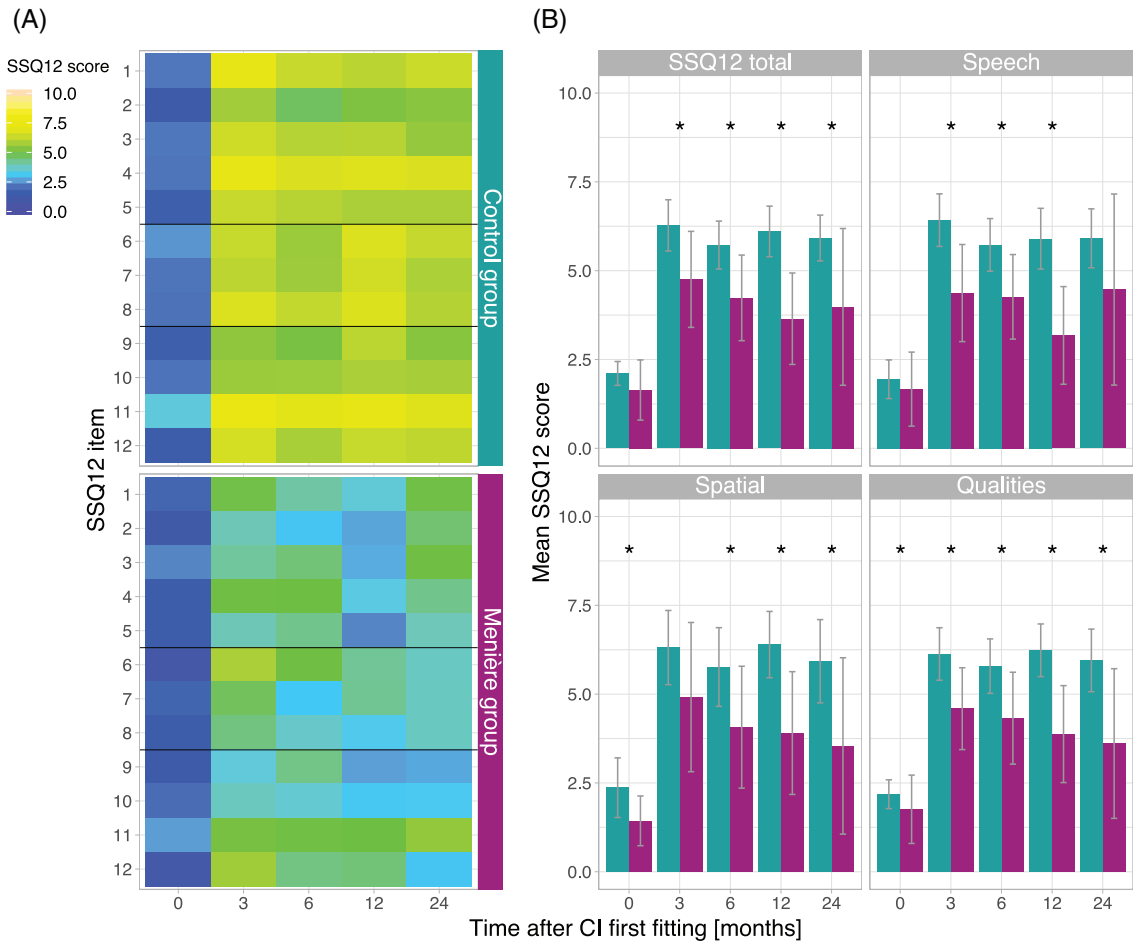

FIGURE 3 Self-assessment of hearing with the *Speech Spatial and Qualities of Hearing Scale* (SSQ12). (A) Heatmap visualizing the cumulative scores by color-coding for single SSQ12-item over the follow-up period of the Menière (purple) and control group (turquoise). (B) Cumulative results for both groups of the postoperative SSQ12 questionnaire in total (upper left), for the category speech (upper right), the category spatial (lower left), and of qualities (lower right). Error bars represent the 95% confidence intervals. Asterisks (*, $p < .05$) indicate significant differences between groups at specific points in time (CI follow-up)

to visualize the distinctly worse self-perceived hearing function of the MD group. The overall SSQ12 mean over time (Figure 3(B) upper left), and broken down for the SSQ12 categories *Speech* (Figure 3(B) upper right), *Spatial* (Figure 3(B) lower left), and *Quality of Hearing* (Figure 3 (B) lower right) demonstrated throughout significantly inferior self-assessed ratings of the MD patients in comparison to the controls. Patients suffering from MD who reported vertigo in the first post-CI year did not significantly differ in SSQ12 scores from MD patients without any vertigo 1 year after implantation (see supplement Figure S1).

4 | DISCUSSION

Since disease duration and progression, as well as age at implantation, have been shown to strongly impact CI outcomes,[11,12] we compared the results of word and sentence tests, as well as the self-perceived hearing function (SSQ12) of MD patients, with a control group consisting of patients best-matched regarding these factors. Results of preoperative best-aided monosyllable testing on the side of implantation were not significantly different, whereas an expected difference was detected in the pure-tone audiometry, with higher thresholds for MD patients at lower frequencies, and vice versa.

The results of the SSQ12 3 months after cochlear implantation revealed considerable increases in scores in both groups, but also significantly lower mean scores of MD patients in comparison to the control group over the time and for all subcategories of the questionnaire. As a more practical short form of the SSQ49, the SSQ12 is a valid instrument to quantify the severity of hearing disabilities.[6–8,10] Outcome measures of the control group in this study are comparable to published data of postlingually severely-to-profoundly hearing impaired, unilaterally and bilaterally cochlear implanted, adult patients,[6,9,13,14] which corroborates the self-perceived underperformance of hearing function in MD patients determined in this study. There are currently few data in the literature of self-perceived hearing function of cochlear implanted MD patients, except for Perkins et al., who reported above-average SSQ12 subscale measures (6 months after CI: speech hearing 7.1 [±1.2], spatial hearing 7.0 [±1.6] and quality of hearing 7.3 [±1.2]) of three individual unilateral MD subjects that underwent cochlear implantation simultaneously to a labyrinthectomy. The sample size is rather small, and the early SSQ12 data 1 month after implantation (initial CI adjustment) is remarkably high (speech hearing 6.1 [±1.9], spatial hearing 6.4 [±0.5] and quality of hearing 6.0 [±1.2]) in comparison to the results of the present study. A recent study by Sanchez-Cuadrado et al. presented very comparable SSQ12 scores of cochlear-implanted MD patients (4.0 [±1.5], $n = 18$) to the present study and showed higher self-assessed hearing function, although not significantly, of their control group (5.0 [±2.0], $n = 18$) as well.[15] Vermeire et al. determined self-assessed hearing function and quality of life in seven cochlear implanted MD patients employing the Nijmegen Cochlear Implant Questionnaire and found poorer results in comparison to other studies of non-MD CI users as well.[16]

In concordance with most of the published studies, which investigated CI outcome performance in MD patients based on standard word and sentence testing,[17–20] subjects of the present study suffering from MD performed equivalently to non-MD patients 1 year after cochlear implantation and later in terms of standard speech testing at 65 dB_{SPL}. This statement must be qualified to some extent, because results of the monosyllable hearing test of this study revealed a significant and sustained underperformance of MD patients in comparison to the controls, when sound intensity was lower, at 55 dB_{SPL}. Comparable data of audiometric testing at lower SPLs is currently lacking in the literature. However, McRacken et al. found significantly poorer monosyllabic word recognition (consonant nucleus consonant test presented at 60 dB_{SPL}) in 21 MD patients compared to a standard sample of 178 adult CI recipients. Interestingly, this was due to the subgroup of 15 non-active MD patients.[21] The authors of the latter study considered neurotoxic effects in the context of MD as possible mechanisms that could worsen CI outcome due to injury to the spiral ganglion neurons (SGN).[21]

Histopathological investigation of temporal bones of MD patients showed endolymphatic lesions as distended membranes (in a non-random manner) throughout the labyrinth,[22] but rather modest structural alterations or changes in the number of SGNs or sensory hair cells (HCs).[23,24] Apart from a decreased number of synapses and afferent endings, ultrastructural evaluation of HCs and SGNs in a patient with unilateral MD revealed smaller axon diameters and of nuclear size of SGNs on the diseased side, as well as in comparison to a cochlea of a longstanding profoundly hearing-impaired, non-MD patient.[25,26] This indication of a neuronal component in MD-pathogenesis is supported by neurotoxic findings in experimental animal models of an endolymphatic hydrops.[27–31] Consequently, SGN pathologies could also potentially limit the performance of electric cochlear stimulation, which, moreover, could then progress in the context of MD.

Among other aspects, the present study is primarily limited by its retrospective design. Prospective studies are needed, but this leads to a second considerable limitation of the present study, the small number of subjects. The prevalence of MD was reported in between 3.5 and 513 per 100.000 cases, and hearing loss typically stabilizes at moderate-to-severe levels; profound hearing impairment or deafness is, therefore, even less frequent.[3,32]

In conclusion, the present group of cochlear-implanted patients suffering from MD reported significantly higher hearing disabilities compared to a matched control group, as measured using the SSQ12. Standardly performed word and sentence tests showed similar results of both groups, but MD patients performed significantly worse when monosyllables were presented at lower sound intensities of 55 db_{SPL}. We think that inferior hearing capabilities at lower SPLs at least partially underlie the self-assessed poorer hearing function in CI-patients of the Menière group as revealed by the SSQ12. Ultrastructural neuronal cochlear injury in the course of MD may be a possible explanation.

MD patients considering a cochlear implantation should be thoroughly informed of a possible underperformance. Long-term prospective, multicenter, studies are needed to achieve more substantial study-subject numbers under equal general conditions to investigate performance and disease progression in cochlear implanted MD patients. This offers the possibility to establish the underlying mechanisms of Meniere's disease.

CONFLICT OF INTEREST

The authors declare no potential conflict of interest.

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SUPPORTING INFORMATION

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