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AUDITORY BRAINSTEM IMPLANTATION AS AN OPTION TO IMPROVE HEARING AND REDUCE TINNITUS: A RETROSPECTIVE STUDY OF FOUR CASES

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Contributions:
A Study design/planning
B Data collection/entry
C Data analysis/statistics
D Data interpretation
E Preparation of manuscript
F Literature analysis/search
G Funds collection

Abstract

Background: Auditory brainstem implantation (ABI) is a modern method of treating hearing impairment, directed especially to patients with neurofibromatosis type 2 (NF2). One of the co-occurring symptoms in these patients is tinnitus; however, little is known about its prevalence and severity. This study evaluated the self-reported hearing ability, hearing-related quality of life, and tinnitus severity in 4 adult ABI users.

Material and methods: The study was a retrospective design. A series of 6 patients who underwent ABI in a single tertiary referral center were asked to fill in two sets of questionnaires referring to their pre- and postoperative experiences. The *Abbreviated Profile of Hearing Aid Benefit* (APHAB) was used to evaluate self-perceived hearing ability and *Nijmegen Cochlear Implant Questionnaire* (NCIQ) to assess hearing-related quality of life. Tinnitus perception was evaluated using *Tinnitus and Hearing Survey* and *Tinnitus Handicap Inventory* (THI).

Results: Of the 6 patients who initially agreed to participate, 4 returned questionnaires. Based on the self-reported results, it was found that each patient noticed an improvement in hearing ability (mean improvement in APHAB score of $M = 25$; $SD = 27$), which was greatest in background noise conditions. A considerable improvement was also noted in the patients' hearing-related quality of life (mean improvement in NCIQ score of $M = 38$; $SD = 13$), which was most pronounced for basic and advanced sound perception. Tinnitus disappeared completely in two patients and was reduced in one patient (mean improvement in THI score for these patients of $M = 25$; $SD = 21$). An increase in tinnitus severity from 26 to 84 points in THI was observed in the remaining patient, who had the greatest intensity of NF2 symptoms in the postoperative period and reported the smallest benefits with ABI.

Conclusion: Brainstem implantation has the potential to improve self-reported hearing ability, hearing-related quality of life, and reduce tinnitus in NF2 patients. However, more prospective studies are needed to confirm and further explore this potential.

Key words: auditory brainstem implantation • tinnitus • hearing • benefits

LOS IMPLANTES AUDITIVOS TRONCALES COMO UNA OPORTUNIDAD PARA MEJORAR LA AUDICIÓN Y REDUCIR EL TINNITUS. ANÁLISIS RETROSPECTIVO DE CUATRO CASOS.

Resumen

Introducción: Los implantes auditivos troncales son un método moderno para tratar la pérdida auditiva, especialmente indicado en pacientes con neurofibromatosis tipo 2. Una de las quejas recurrentes en estos pacientes es el tinnitus. Sin embargo, se sabe poco sobre su prevalencia y gravedad. El estudio evaluó el impacto de la implantación troncal en la audición, la calidad de vida y la molestia del tinnitus en 4 pacientes sometidos a la implantación troncal.

Material y métodos: Estudio retrospectivo, se analizaron seis pacientes que se sometieron a la implantación del tronco en un mismo centro de tercer nivel. Se les pidió que completaran dos conjuntos de cuestionarios sobre sus experiencias pre y postoperatorias. El cuestionario APHAB se utilizó para evaluar la capacidad auditiva subjetiva y el cuestionario NCIQ para evaluar la calidad de vida relacionada con la audición. La percepción del tinnitus se evaluó mediante cuestionarios THS y THI.

Resultados: De los 6 pacientes que inicialmente aceptaron participar en el estudio, 4 respondieron los cuestionarios. Basado en los resultados, se encontró que cada paciente informó una mejora en la capacidad auditiva (mejora promedio en los cuestionarios APHAB con el valor *M* = 25; *SD* = 27), que fue el más alto en las condiciones de ruido de fondo. También se observó una mejora significativa en la calidad de vida de los pacientes relacionados con la audición (mejoría promedio en la escala NCIQ *M* = 38; *SD* = 13), que fue más pronunciada en el caso de la percepción del sonido básica y avanzada. El tinnitus desapareció por completo en dos pacientes y disminuyó en un paciente (mejora media en la puntuación de THI para estos pacientes en *M* = 25; *SD* = 21). Se observó un aumento en el tinnitus de 26 a 84 puntos en THI en el último paciente que tuvo los síntomas más graves de NF2 en el postoperatorio y tuvo el menor beneficio de ABI.

Conclusión: La implantación troncal puede mejorar la capacidad auditiva subjetiva, la calidad de vida asociada con la audición y reducir la gravedad del tinnitus en pacientes con NF2. Sin embargo, se necesita más investigación en estudios prospectivos para confirmar su efectividad y explorar más a fondo este potencial.

Palabras clave: implantes auditivos troncales; tinnitus; audición; beneficios

СЛУХОВЫЕ СТВОЛОМОЗГОВЫЕ ИМПЛАНТЫ – ВОЗМОЖНОСТЬ УЛУЧШЕНИЯ СЛУХА И УМЕНЬШЕНИЯ ШУМА В УШАХ. РЕТРОСПЕКТИВНЫЙ АНАЛИЗ ЧЕТЫРЕХ СЛУЧАЕВ

Аннотация

Введение: Слуховая стволомозговая имплантация является современным методом лечения тугоухости, предназначенным в особенности для пациентов с нейрофиброматозом II типа. Шум в ушах является сопутствующей жалобой у таких пациентов. Тем не менее, существует мало информации на тему распространенности данного недуга и его степени тяжести. В данном исследовании была проведена оценка влияния стволомозговой имплантации на слух, качество жизни и шум в ушах у 4 пациентов, которым был вживлен слуховой стволомозговой имплант.

Материалы и методы: Исследование имеет ретроспективный характер. Шесть пациентов, после стволомозговой имплантации, которая была проведена в одном и том же центре с 3-й степенью референтности, получили два набора анкет с вопросами, касающимися периода перед и после операции. Для субъективной оценки слуха была использована анкета APHAB. Анкета NCIQ использовалась для оценки качества жизни, связанного со слухом. Восприятие шума в ушах оценивалось с помощью опросников THS и THI.

Результаты: Из 6 пациентов, которые изначально согласились принять участие в исследовании, 4 вернули заполненные вопросники. На основании полученных результатов было обнаружено, что каждый пациент отметил улучшение слуховой способности (среднее улучшение в опросниках APHAB: *M* = 25; *SD* = 27), особенно в условиях фонового шума. Значительное улучшение было также отмечено в качестве жизни пациентов, связанного со слухом (среднее улучшение по шкале NCIQ: *M* = 38; *SD* = 13), которое было наиболее выраженным в случае основного и расширенного восприятия звука. Шум в ушах полностью исчез у двух пациентов и снизился у одного пациента (среднее улучшение показателей THI для этих пациентов: *M* = 25; *SD* = 21). Увеличение шума в ушах с 26 до 84 баллов в THI наблюдалось у последнего пациента, который имел наиболее выраженные симптомы NF2 в послеоперационном периоде и отметил наименьшую эффективность стволомозговой имплантации.

Выводы: Стволомозговая имплантация может улучшить субъективную слуховую способность, качество жизни, связанное со слухом, и снизить степень шума в ушах у пациентов с NF2. Тем не менее, необходимы дополнительные проспективные исследования, чтобы подтвердить эффективность и потенциал данного вида имплантации.

Ключевые слова: слуховые стволомозговые импланты • шум в ушах • слух • эффекты

SŁUCHOWE IMPLANTY PNIOWE JAKO SZANSA NA POPRAWĘ SŁYSZENIA I REDUKCJĘ SZUMÓW USZNYCH – RETROSPEKTYWNA ANALIZA CZTERECH PRZYPADKÓW

Streszczenie

Wstęp: Wszczepianie słuchowych implantów pniowych jest nowoczesną metodą leczenia niedosłuchu, przeznaczoną szczególnie dla pacjentów z neurofibromatozą typu 2. Jedną ze współwystępujących dolegliwości u tych pacjentów są szumy uszne, jednak niewiele wiadomo na temat ich występowania i ciężkości. W badaniu oceniono wpływ implantacji pniowej na słyszenie, jakość życia oraz uciążliwość szumów usznych u 4 pacjentów poddanych zabiegowi implantacji pniowej.

Materiał i metody: Badanie miało charakter retrospektywny. Sześcioro pacjentów, którzy zostali poddani implantacji pniowej w tym samym ośrodku o 3. stopniu referencyjności, poproszono o wypełnienie dwóch zestawów kwestionariuszy odnoszących się do ich doświadczeń przed- i pooperacyjnych. Kwestionariusz APHAB został wykorzystany do oceny subiektywnej zdolności słyszenia, a kwestionariusz NCIQ – do oceny jakości życia związanej ze słuchem. Percepcję szumów usznych oceniano za pomocą kwestionariuszy THS oraz THI.

Wyniki: Spośród 6 pacjentów, którzy początkowo zgodzili się wziąć udział w badaniu, 4 zwróciło kwestionariusze. Na podstawie wyników stwierdzono, że każdy pacjent odnotował poprawę zdolności słyszenia (średnia poprawa wyniku w kwestionariuszy APHAB o wartości *M* = 25; *SD* = 27), która była największa w warunkach hałasu tła. Znaczną poprawę odnotowano także w zakresie jakości życia pacjentów związanej ze słuchem (średnia poprawa w skali NCIQ *M* = 38; *SD* = 13), co było najbardziej wyraźne w przypadku podstawowej i zaawansowanej

percepcji dźwięku. Szumy uszne zniknęły całkowicie u 2 pacjentów i zmniejszyły się u 1 pacjenta (średnia poprawa wyniku THI dla tych pacjentów o $M = 25$; $SD = 21$). Wzrost nasilenia szumów usznych z 26 do 84 punktów w THI zaobserwowano u ostatniego pacjenta, który miał największe nasilenie objawów NF2 w okresie pooperacyjnym i odnotował najmniejsze korzyści z ABI.

Wnioski: Implantacja pniowa może poprawić subiektywną zdolność słyszenia, jakość życia związaną ze słyszeniem i zredukować uciążliwość szumów usznych u pacjentów z NF2. Potrzeba jednak większej liczby badań prospektywnych w celu potwierdzenia ich skuteczności i potencjału.

Słowa kluczowe: słuchowe implanty pniowe • szumy uszne • słyszenie • korzyści

Abbreviations

ABI – auditory brainstem implant
APHAB – *Abbreviated Profile of Hearing Aid Benefit*
NCIQ – *Nijmegen Cochlear Implant Questionnaire*
NF2 – neurofibromatosis type 2
THI – *Tinnitus Handicap Inventory*
THS – *Tinnitus and Hearing Survey*

Background

The use of auditory implants has allowed for effective auditory rehabilitation in a number of patients with various types of hearing losses. The most commonly used implantable hearing prostheses are cochlear implants. In Poland, the national program of treating hearing loss with the help of cochlear implants was initiated by the team of prof. Henryk Skarzynski in 1992 [1]. Since that time, there has been a dynamic development of both surgical techniques and fitting methods [2] that allow the best possible hearing preservation and speech understanding [3–5].

However, in some cases it is impossible to successfully use a cochlear implant due to disturbed transmission in the auditory nerve. A solution in this situation might be an auditory brainstem implant (ABI), a modern method of treating hearing loss caused by damage to the auditory nerve(s). It is intended mainly for patients diagnosed with neurofibromatosis type 2 (NF2). NF2 is a genetic disease with autosomal dominant inheritance. In the clinical picture, skin and ocular neoplastic lesions are observed. Its other characteristic feature is the occurrence of bilateral tumors of the vestibular part of the auditory nerve (called schwannomas). These can lead to progressive retrocochlear hearing loss, dizziness, and headaches as well as troublesome tinnitus [6–8].

ABI surgery relies on placing a prosthesis that electrically stimulates the brainstem. It is a complicated procedure and requires the cooperation of many specialists. In Poland, the first implantation of a brainstem implant was performed in 1998 [9]. Very good results obtained with the first patient has allowed the treatment of hearing loss with this method to continue with subsequent patients. The primary purpose of auditory implantation is to improve speech intelligibility. However, studies conducted among patients with cochlear implants indicate that the reduction of annoying tinnitus is also important from the perspective of patients' quality of life [10,11].

Tinnitus is one of the most common symptoms associated with hearing loss in patients qualified for auditory implantation. It is defined as a phantom sound sensation, i.e. sound without the presence of an external acoustic source. Tinnitus may lead to disruption in the daily functioning of the patient [12,13]. To the authors' best knowledge, the first to describe the suppression effect of ABI on tinnitus were Soussi and Otto [14] in 1994. In their work, the authors found that the daily use of ABI led to a tinnitus reduction in 85.7% of evaluated patients; however, no validated measures of tinnitus severity or handicap were used. Some 25 years later, only a few papers have been published on tinnitus reduction from the use of a brainstem implant. A comprehensive report has recently been published by the same center [15]. Based on a validated tinnitus questionnaire, the authors found additional evidence supporting the assumption that tinnitus is suppressed after ABI. In their study, ABI users with NF2 reported significant tinnitus handicap reduction from wearing the device.

The aim of the present study is to add new evidence relevant to this surprising finding by assessing the effect of brainstem implantation on hearing under various acoustic conditions, the health-related quality of life, and the reduction of tinnitus in Polish patients. Based on the available literature, the authors hypothesize that a brainstem implant not only improves hearing and health-related quality of life, but also reduces tinnitus.

Material and methods

This retrospective study was conducted among a group of patients with a brainstem implant under the care of the Institute of Physiology and Pathology of Hearing (IPPH, Warsaw, Poland). After obtaining initial interest in participating in the study from the patients via a phone call, two sets of questionnaires were sent to them by post: one relating to the pre-implantation period, where patients were asked to retrospectively evaluate their hearing under different acoustic conditions, health-related quality of life, and tinnitus severity; the second set was the same questionnaire but the patients were asked to make an assessment based on their current perception. Both sets of questionnaires were sent at the same time. The study procedures complied with the Declaration of Helsinki.

Surgery

The ABI surgeries of 3 patients were carried out by an experienced team of oto- and neurosurgeons at IPPH. One patient (Patient 2) was operated on at another center (abroad) but all the auditory rehabilitation was done at our center. All patients received the Med-El ABI (Innsbruck, Austria), which consists of two components: an external audio processor and an internal implant with a 12-electrode array. Operations were conducted using a retrosigmoid approach. A number of electrophysiological tests were performed to decide on the optimal placement of the ABI electrodes. Electrically evoked auditory brainstem responses were intraoperatively recorded to ensure that the electrodes activated the auditory system and did not stimulate nearby nonauditory structures. The electrode

array was then introduced, placing it in the area vestibularis, also under electrophysiological control.

Self-report measures

The assessment of subjective hearing in different acoustic situations was made on the basis of the widely used *Abbreviated Profile of Hearing Aid Benefit* (APHAB) [16]. The questionnaire consists of 24 questions aimed at assessing hearing benefits obtained with a hearing aid in the following areas: Ease of Communication, Background Noise, and Reverberation; another scale is designed to measure Aversiveness to sound. The higher the APHAB score, the less the subject's hearing benefits.

The hearing-related quality of life was tested using the *Nijmegen Cochlear Implant Questionnaire* (NCIQ) [17], which is intended mainly for cochlear implant users. In the current study, an experimental version of the tool was used to meet the needs of ABI users. The NCIQ consists of 60 questions assigned to six subscales: Basic Sound Perception, Advanced Sound Perception, Speech Production, Psychological, Activity Limitations, and Social Interactions. The higher the NCIQ score, the better the hearing-related quality of life.

Self-reported tinnitus severity was measured with the *Tinnitus Handicap Inventory* (THI) [18–20]. This questionnaire assesses the general handicap caused by tinnitus and is one of the most frequently used questionnaires for gauging the effectiveness of tinnitus treatment. The questionnaire consists of 25 items. The higher the THI score, the higher the tinnitus severity. Additionally, in order to differentiate which of the symptoms – hearing loss or tinnitus – was a bigger problem for the patient, the *Tinnitus and Hearing Survey* (THS) [21,22] was used. This questionnaire is a screening tool designed to differentiate the effects of tinnitus and those of hearing loss. THS consists of 10 statements and is divided into three main parts: A, Tinnitus; B, Hearing; and C, Sound Tolerance. The higher the THS score, the bigger the problem caused by the evaluated symptom.

Patients were also asked to fill in a form constructed by the study authors in which they were asked to provide gender, age, time of becoming an ABI user, subjective benefits with ABI (on a 5-level *Likert scale* from 1, no benefits, to 5, very big benefits), and the localization and nature of the tinnitus.

Results

Because NF2 is characterized by a high percentage of deaths [23], one patient (among the 7 found in the medical charts of IPPH) died before the study began. Questionnaires were sent to the remaining 6 patients, 4 of which were returned (a rate of 67%).

The characteristics of the study participants are presented in Table 1. The participants were adults aged from 29 to 42 years at the time of the study. They were predominantly female (75%). Patients used their ABI at least 12 hours a day and in most cases reported large subjective benefits with the device.

Subjective change in hearing and health-related quality of life

A subjective assessment of the patients' hearing in different acoustic conditions based on APHAB is presented in Figure 1. Mean improvement in APHAB score was $M = 24.7$; $SD = 26.5$. Each patient noticed an improvement in hearing; the greatest benefits were reported under the background noise condition. In two of four patients an increased aversiveness to sound was noted. The smallest benefits were observed in Patient 3. Based on the medical interview, the highest progression of NF2 symptoms was noted in this patient, which significantly impaired her overall health.

Results of the NCIQ are presented in Figure 2. Mean improvement in NCIQ score was $M = 37.7$; $SD = 13.25$, which was most pronounced for basic and advanced sound perception. In two of the four patients a considerable improvement in health-related quality of life was noted. A smaller improvement was observed in Patient 4. Once again, due to the general deterioration of health status in Patient 3, her hearing-related quality of life was only slight.

Subjective change in tinnitus handicap

Tinnitus characteristics. All patients reported tinnitus prior to surgery. Patient 1 described the tinnitus as high sounds comparable to a kettle squeak, moderately loud, and moderately bothersome. After the operation tinnitus disappeared completely in the right ear; however, it remained in the left ear. Patient 2 complained of moderately bothersome, monotonous tones and noises, of

Table 1. Patient characteristics

Patient no.	Gender	Age	Operated side	Time of being an ABI user	ABI use per day (hours)	Subjective benefit with ABI	PTA* preop	PTA* postop
1	F	29	R	3 years	17	+	120	120
2	M	31	R	10 years	12	+	114.4	120
3	F	37	L	3 years	15	±	85	118.3
4	F	42	L	3 years	13	+	56.6	120

F, female; M, male; R, right; L, left; + patient reported subjective benefit with ABI; ± patient reported moderate benefit with ABI; * PTA, pure tone average (mean of hearing levels for octave frequencies of 125–8000 Hz; for no response at max output (120 dB), the max value was used)

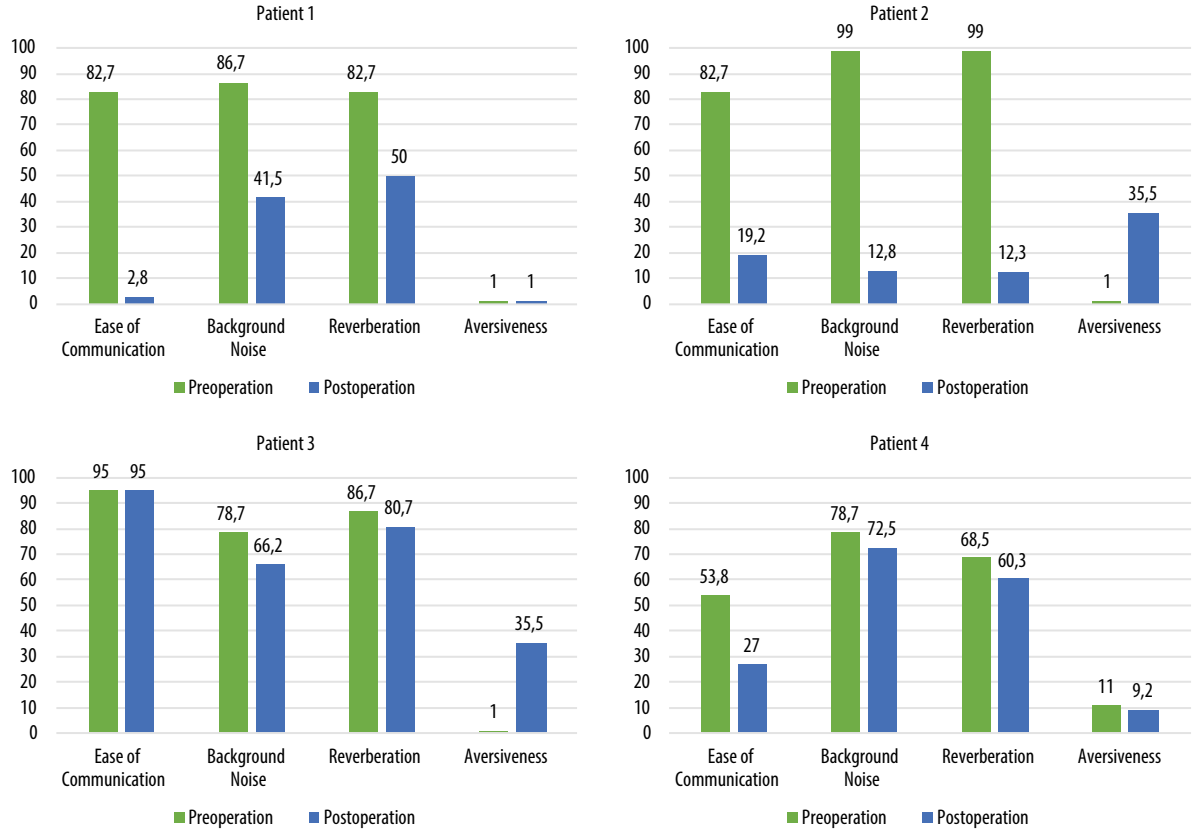

Figure 1. Pre- and postoperative APHAB results of individual patients.

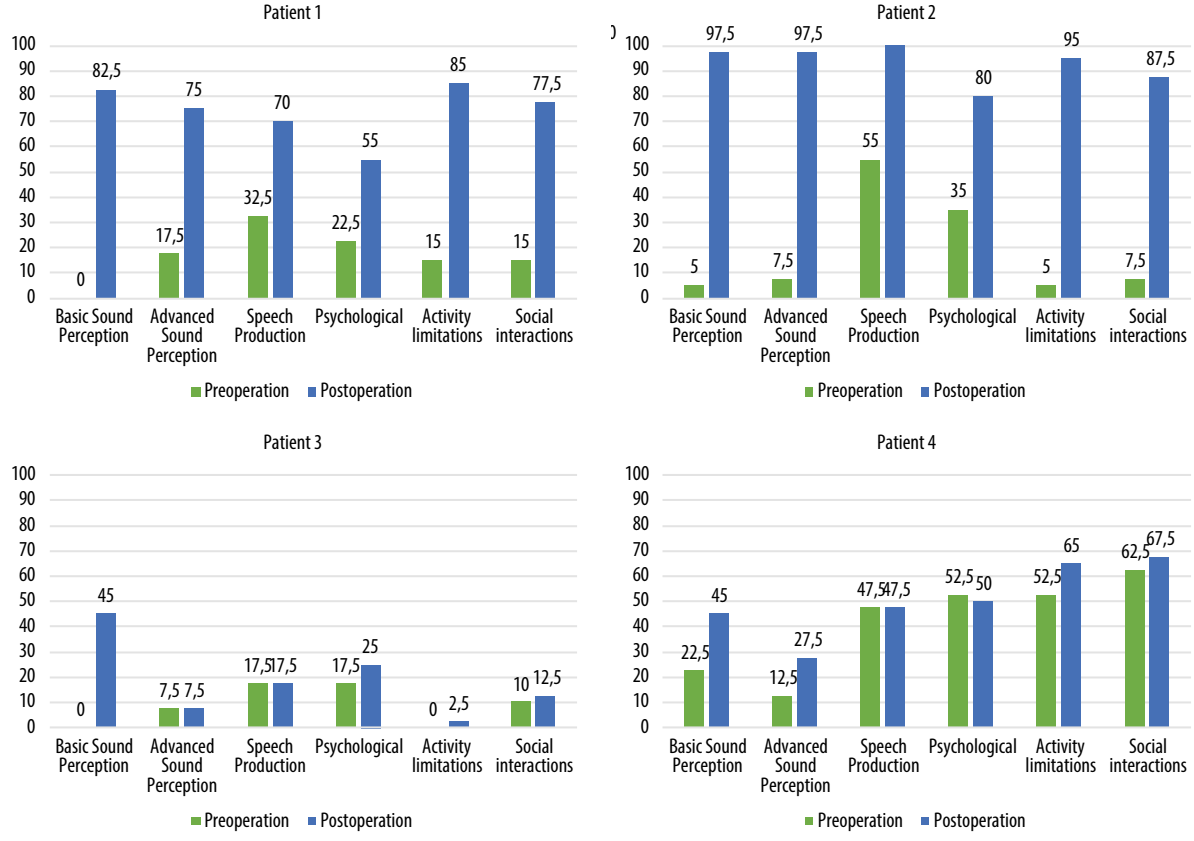

Figure 2. Pre- and postoperative NCIQ results of individual patients.

medium high, quite loud, like a crackling or chirp. After ABI surgery, the tinnitus remained binaural, both when the speech processor was in use and after its removal. Patient 3 described her tinnitus as a simultaneous tone and noise, medium high, not too loud, but troublesome. After the ABI, tinnitus was present only in the left (not operated) ear, both when wearing the speech processor and after removing it. Patient 4 complained of a monotonous tinnitus tone, medium high, moderately loud, and moderately troublesome. After the operation, the patient experienced tinnitus only in the right (not operated) ear, both when wearing the speech processor and after removing it.

Questionnaire results. The results of the THI and THS questionnaires are presented in Figure 3 and 4, respectively. In all patients, hearing problems were greater than tinnitus problems. Based on the results, it was found that for most patients the ABI treatment not only improved hearing ability, but also contributed to a reduction of tinnitus. Mean improvement in THI score was $M = 25$; $SD = 21$.

Discussion

To date, few tinnitus studies have been published on patients who have undergone brainstem implantation. More papers have been concerned with assessing hearing parameters [24–26], which indicate a clinically significant improvement in hearing ability. These findings are in line with our study, where an improvement in hearing ability and health-related quality of life was reported by all evaluated ABI users.

Although the majority of patients in our study reported that hearing problems were more pronounced than tinnitus problems, tinnitus severity was assessed by them as mild to moderate, indicating some disruptions in everyday functioning. Based on our results, we noted that ABI also has the potential to reduce tinnitus, possibly in a similar way to other implantable hearing prostheses such as cochlear implants [27], middle ear implants [28], or bone-conduction implants [29]. However, there are still patients who complained about tinnitus after surgery.

There are many hypotheses as to why not all patients achieve the expected improvement in hearing parameters and tinnitus reduction after ABI surgery. One of them is that the intensity of the symptoms may relate to the severity of pathological changes caused by advancement of the underlying disease. Additionally, the lack of appropriate rehabilitation and postoperative complications (such as, for example, the displacement of electrodes or problems with the choice of speech processor) can also negatively affect the postoperative results [30].

Tinnitus suppression mechanisms in ABI users

The exact mechanism explaining tinnitus suppression in ABI users is not fully known. In the literature, two possible hypotheses regarding tinnitus suppression by auditory implants have been discussed [15].

The first assumes that tinnitus suppression is caused by masking, an idea which is supported by both early [31] and more recent [32] studies on cochlear implant users, who additionally benefited from some kind of sound therapy (from hearing aids or a dedicated app) for their tinnitus suppression. A masking theory has also been partially supported by Roberts et al. [15] who found that ABI users reported tinnitus suppression immediately after the device was activated, but the effect did not persist longer than an hour after the device was deactivated.

The second theory explaining the tinnitus suppression in ABI users is that the device has a direct effect on the brain or brainstem. For example, Argence et al. [33] found in their experimental study on rats that some receptors in the central nucleus of the inferior colliculus can, due to the electrophysiological stimulation, be re-regulated and in this way suppress tinnitus perception.

In recent years interesting neural tinnitus suppression models have been proposed, relevant also to human subjects [34–36]. It is worth noting that 3 of our 4 patients reported that after ABI surgery tinnitus completely disappeared on their operated side, while persisting on the other, non-operated side. Roberts et al. [15] found that 39.1% of their patients claimed that tinnitus suppression lasted also when the ABI was off.

Taking into account that tinnitus is a multifactorial symptom, depending on a range of psychological and sociodemographic factors [37,38], further studies exploring the mechanisms that alter its perception are needed, as in auditory implant users.

Limitations of the study and future directions

Apart from certain advantages, our study also has limitations. First of all, it was a retrospective design and so was prone to recall bias – especially taking into consideration that two sets of questionnaires were sent to patients at the same time. It is known that people are more likely to remember certain events, or exaggerate or minimize some symptoms after time [39], which could affect our results. Additionally, none of the questionnaires used in the current study was created specifically for the needs of ABI users, which might question their validity and reliability in this particular group of patients [40]. Also, questionnaire responders – in comparison to non-responders – are generally more motivated and healthier [39]. It is not known what the results of our examination would have been if all 6 people contacted by phone had responded to the questionnaires, especially considering the large number of questions and the low number of participants. Another disadvantage of retrospective case series is that they are uncontrolled [41] – we did not have the opportunity to compare the hearing benefits and tinnitus severity of our participants with other NF2 patients who were not implanted with an ABI.

In future it is recommended that prospective studies on a bigger groups of ABI users be conducted to confirm the results obtained in the current study. A rigorous methodology and adequate sample size is necessary to decide on an intervention's effectiveness [42]. Currently, only a general association, and not causation [43], can be inferred from the studies on the hearing benefits and tinnitus improvement in ABI users.

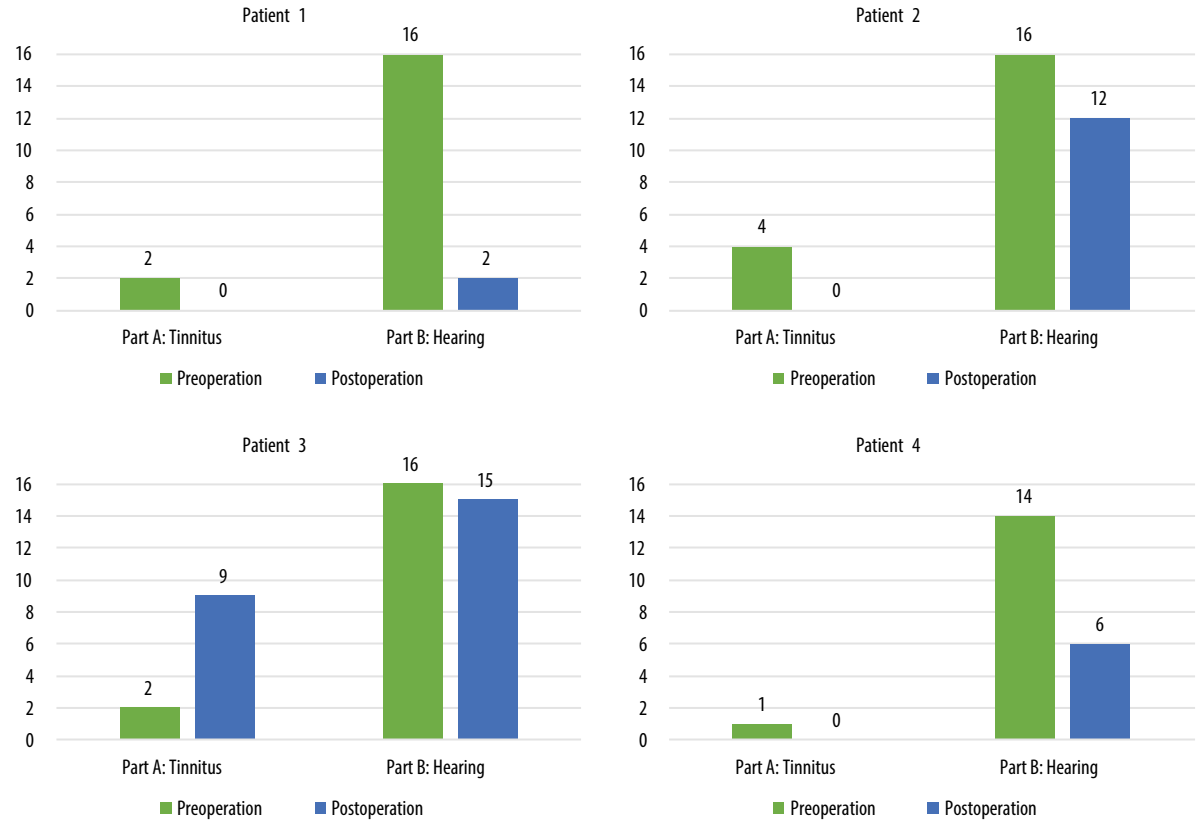

Figure 3. Pre- and postoperative THS results of individual patients.

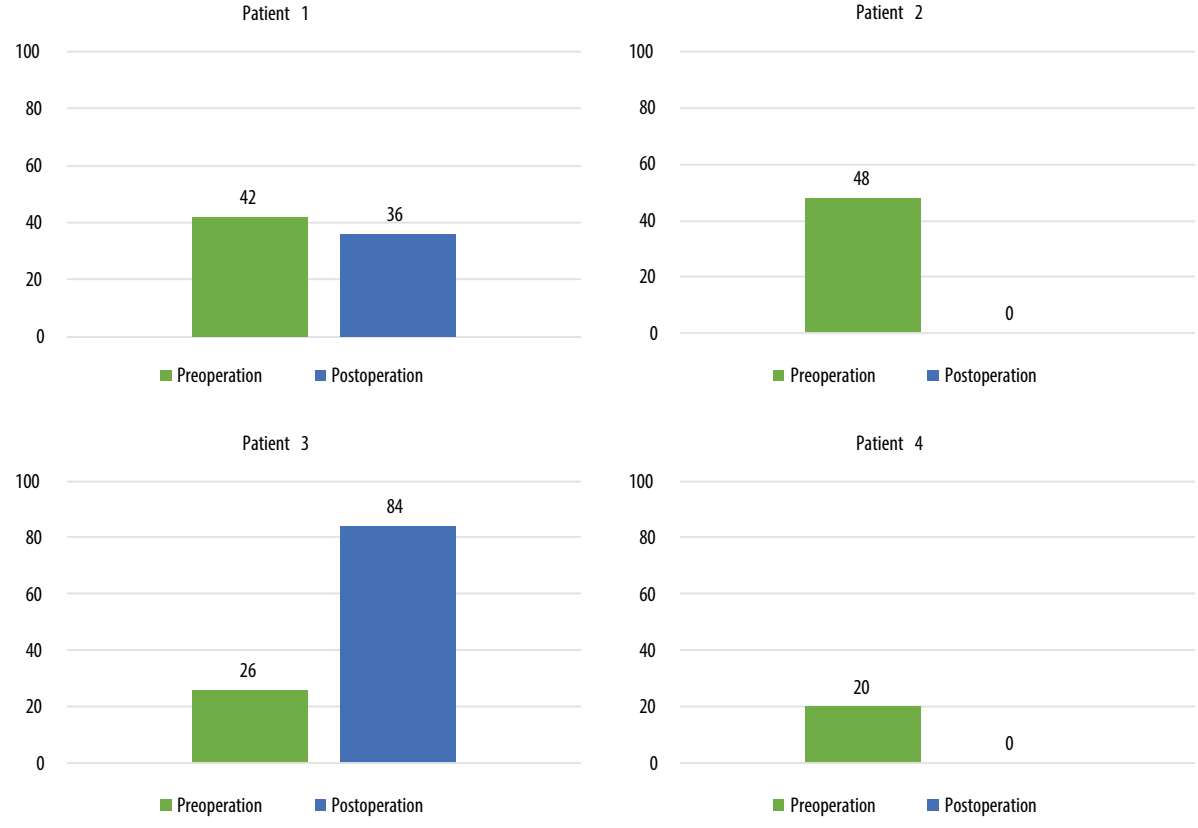

Figure 4. Pre- and postoperative THI results of individual patients.

From a clinical point of view, it also needs to the highlighted that brainstem implantation is suitable only for a select group of patients suffering from NF2. The size of the tumor affecting the cochlear nuclei and its vascularization may be contraindications for surgery. The location of the electrodes during the operation and during activation of the device is also important, requiring appropriate anatomical conditions. All of these factors can affect the course of surgery and the postoperative improvements obtainable.

Conclusions

Brainstem implantation has the potential to improve self-reported hearing ability, hearing-related quality of life, and reduce tinnitus in patients with NF2. However, more prospective studies are needed to confirm and further explore this potential.

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