Otology

Benefits of bone conduction hearing aid in children with unilateral aural atresia

Beneficio dell'apparecchio acustico a conduzione ossea nei bambini con atresia auris unilaterale

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SUMMARY

Objective. To assess the hearing benefit with a unilateral bone conduction hearing aid in a cohort of children with unilateral aural atresia.

Methods. Cross-sectional case series pilot study involving 7 children (median age: 10 years, range 6-11). All patients underwent pure-tone, speech, aided sound field and aided speech audiometry and Simplified Italian Matrix Test (SIMT) with and without bone conduction hearing aid (Baha 5® Cochlear™). Cognitive abilities were assessed in 5 patients.

Results. The mean air conduction pure-tone average (PTA) of the atretic ear was 63.2 ± 6.9 dB, while the bone conduction PTA was 12.6 ± 4.7 dB. Speech discrimination score of the atretic ear was 88.6 ± 3.8 dB, while with the hearing aid it was 52.8 ± 1.9 dB. In the contralateral ear, there was no significant air-bone gap, and PTAs for air and bone conduction thresholds were within normal range (PTA ≤ 25 dB). The mean aided air conduction hearing threshold was 26.2 ± 7.97. Mean speech recognition threshold without the hearing aid was -5.1 ± 1.9 dB, and -6.0 ± 1.7 dB with the hearing aid tested with the SIMT. The mean score of the cognitive test was 46.8 ± 42.8.

Conclusions. These preliminary findings should encourage clinicians in proposing a unilateral bone conduction hearing aid in children with unilateral atresia.

KEY WORDS: aural atresia, bone conduction hearing aids, unilateral hearing loss, conductive hearing loss, Simplified Italian Matrix Test

RIASSUNTO

Obiettivo. *Verificare il beneficio uditivo con protesi monolaterale per via ossea in pazienti affetti da atresia auricolare unilaterale, una rara causa di ipoacusia trasmissiva unilaterale congenita.*

Metodi. *Uno studio pilota trasversale di casi ha coinvolto 7 bambini (età mediana di 10 anni, intervallo 6-11). I pazienti sono stati sottoposti ad audiometria tonale, vocale, tonale protesica e vocale protesica e Matrix test Italiano semplificato (SIMT) con e senza apparecchio acustico a conduzione ossea (Baha 5® CochlearTM).*

Risultati. *La soglia tonale media (PTA) della conduzione aerea dell'orecchio atresico era di 63,2 ± 6,9 dB, mentre la PTA della conduzione ossea era di 12,6 ± 4,7 dB. Il punteggio di discriminazione vocale (SDS) dell'orecchio atretico era di 88,6 ± 3,8 dB, con l'apparecchio acustico l'SDS era di 52,8 ± 1,9 dB. Nell'orecchio controlaterale, non vi era alcun gap aereo-osso significativo e le PTA per le soglie di conduzione aerea e ossea erano all'interno dell'intervallo normale (PTA ≤ 25 dB). La soglia uditiva media per via aerea con l'apparecchio acustico era di 26,2 ± 7,97 dB. La soglia media di riconoscimento vocale senza l'apparecchio acustico era -5,1 ± 1,9 dB, mentre era -6,0 ± 1,7 dB con l'apparecchio acustico testato con il SIMT. Il punteggio medio del test cognitivo era 46,8 ± 42,8.*

Conclusioni. *I risultati preliminari di questo studio incoraggiano i medici a proporre l'uso unilaterale di un apparecchio acustico a conduzione ossea nell'atresia unilaterale nei bambini.*

PAROLE CHIAVE: *atresia auricolare, protesi acustiche a conduzione ossea, ipoacusia unilaterale, ipoacusia trasmissiva, Matrix Test Italiano Semplificato*

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Introduction

Unilateral aural atresia is a rare condition in which the external auditory canal is not developed thus causing a pure conductive hearing loss up to around 50 dB in air-bone gap [1]. Aural atresia is considered part of the oculo-auriculo-vertebral spectrum (OAVS), a rare syndrome [2] in which patients can present a severe phenotype with aural, facial, ocular and cervical spinal involvement called Goldenhar syndrome. The craniofacial structures derived from the first and second pharyngeal arches may be altered on one (85% of cases) or both sides [3-5]. The resulting typical facial asymmetry [6] might be accompanied by anomalies involving cranial nerves [7], brain, internal carotid artery, inner ear [8] and salivary glands [9]. These multiple anomalies can have different clinical consequences, and in particular the hearing loss caused by the aural atresia can have an impact on the communication and learning skills during the development. Although in bilateral aural atresia hearing rehabilitation with bone conduction hearing aids is nowadays mandatory with a specific bilateral fitting [10], in the latest years the necessity to rehabilitate a unilateral deficit has also emerged [11]. Indeed, unilateral hearing loss impacts sound localisation and speech perception in background noise, thus being a non-negligible factor in the hearing and speech evolution of the child [12-14]. Unfortunately, most of the literature on this topic is based on small studies [15,16]. Nevertheless, recent studies show that patients affected by unilateral hearing loss may present poor academic [17] and speech performance [18], with non-negligible impact on the quality of life of the children [19]. Patients with unilateral atresia can face ominous difficulties in noisy environments, with multiple speakers and high cognitive load, and possible impact on scholastic performance causing long term disabilities. This represents a major issue if concomitant cognitive impairment is present. Nowadays, the currently available bone conduction hearing aids are small, light and interconnected with smartphones, tablets, or remote microphones, which are extremely useful for paediatric patients, especially in scholastic environments in which background noise can impair listening abilities.

Up to now, some useful tests to prove hearing performance in background noise are available, such as the Simplified Italian Matrix Test (SIMT) that can give a quantitative evaluation of the hearing impairment in noisy conditions [20]. Hearing benefit with bone conduction hearing aids is reported in conductive hearing loss due to multiple aetiologies such as chronic otitis, cholesteatoma and otosclerosis [21] in adult patients, but studies on the possible benefits in background noise in the paediatric population with unilateral aural atresia are still lacking.

To evaluate the possible benefit of a bone-conduction hearing aid (BCHA) in background noise, we performed an audiological study on a group of paediatric patients with unilateral aural atresia using the SIMT.

Materials and methods

Patients

Seven consecutive children (3 females and 4 males, mean age 9 ± 2 years, median 10, range 6-11) presenting with congenital unilateral aural atresia underwent audiological evaluation with and without bone conduction hearing aid in the affected side at a third referral University centre. Five patients presented unilateral atresia on the right side. Five patients underwent evaluation of cognitive abilities.

No patient had previous experience of hearing aids according to the parents' choice, despite being informed since the first phases of the child's life. All patients presented appropriate development according to speech and language evaluations.

Audiological assessment

The evaluation of hearing performance was carried out by clinicians and technicians experienced in the management of hearing loss and rare congenital diseases. Audiological assessment included evaluation of medical history and otoscopy, pure-tone, speech, aided sound field and aided speech audiometry and SIMT. All tests were conducted in a 4.2 m x 2.5 m soundproof booth, using an Otometrics MADSEN® Astera2 audiometer with a set of TDH39 headphones and Indiana Line loudspeakers.

Pure tone audiometry (PTA) was performed for both air and bone conduction and the PTA was measured for the frequencies 0.5, 1, 2 and 4 kHz. Aided sound field audiometry was performed with the bone conduction hearing aid positioned on the mastoid of the affected ear by means of a headband. On the contralateral normal hearing ear, masking narrow band noise was administered with headphones at 50 dB over the hearing threshold.

Speech audiometry was performed in quiet, testing the atretic side with headphones, administering masking white noise in the contralateral normal ear. Aided speech audiometry was performed in free field conditions, with the bone conduction hearing aid positioned on the mastoid of the affected ear using a headband. On the contralateral normal hearing ear, masking white noise was administered by means of headphones. Speech discrimination score (SDS) represents the sound level (in dB) at which the patient correctly identifies 100% of administered words.

The Italian Matrix Sentence Test (IMST) is a speech audiometry in background noise with automatically adapted signal to

noise ratio (SNR). It is structured into 20 randomised lists of five-word sentences, semantically unpredictable and administered after a training session to minimise the learning curve [22]. The SIMT is specific for children aged 4 years or more and uses 14 randomised lists of three-word sentences with the same structure (number, adjective, noun). The background noise is presented at a fixed 65 dB sound pressure level (SPL), while the speech material is actively adapted by the software. The result of the test is a score representing the SNR in dB at which the patient can recognise 50% of the speech material: the speech recognition threshold (SRT) [20].
A specific setup finalised to reproduce as faithfully a classroom environment was chosen. Two loudspeakers were placed respectively 1 meter in front and behind the subject, the noise was presented at a fixed level of 65 dB at 0° and 180° azimuth, the speech was presented at 0° azimuth (Fig. 1) and the sentence level was automatically adapted according to the preceding response.

Hearing aid

All patients were evaluated with and without the same bone conduction hearing aid. The chosen hearing aid was the Baha 5® Cochlear™ set for a normal bone conduction threshold (PTA under 25 dB nHL). The positioning of the hearing aid was performed by a physician experienced in audiology using a headband. Children were invited to wear the device for at least an hour after which they underwent audiological testing. The correct position of the Baha 5® Cochlear™ was checked by a technician right before testing.

Assessment of cognitive abilities

Patients underwent evaluation of mental abilities performed with a developmental psychologist experienced in paediatric development and hearing loss. The evaluation was performed after the audiological evaluation to maximise children's attention capacities during the audiological testing. Two of the children were too tired for further activities and results of the cognitive tests were not considered reliable.
Patients were evaluated with the Raven's Colored Progressive Matrices (CPM) test [1], the leading global non-verbal measure of mental ability in research, educational and clinical settings. The CPM test comprises 36 items (brightly coloured to attract and maintain children's attention), divided into three sets of 12 (set A, Ab and B), and ordered in terms of increasing difficulty. The sets have been designed to distinguish between degrees of intellectual maturity by quantifying a child's ability to form comparisons and to reason by analogy [23].
The Raven's CPM results in a score that can be converted into a percentile, by mean of a specific conversion table also considering the age of the subjects.
Patients scoring less than 5 are considered to have mental abilities strongly below average, between 5 and 25 with results below average, results between 25 and 75 are considered normal, scoring higher than 75 is considered above average and more than 95 are considered strongly above average.

Statistical analysis

Continuous variables were expressed as median and range. The nonparametric Wilcoxon Signed Rank Sum test was used to assess the modifications in hearing status before and after Baha 5® Cochlear™ use. Spearman test was applied to identify correlations between continuous variables. The level of significance was $\alpha = 0.05$. Statistical analysis was carried out with SPSS 24.0 software.

Results

Audiological and cognitive evaluation results for each patient are presented in Table I. The median air conduction hearing threshold of the atretic ear, expressed as PTA was 66.2 dB nHL (range 28.7-70.0 dB nHL), while the median

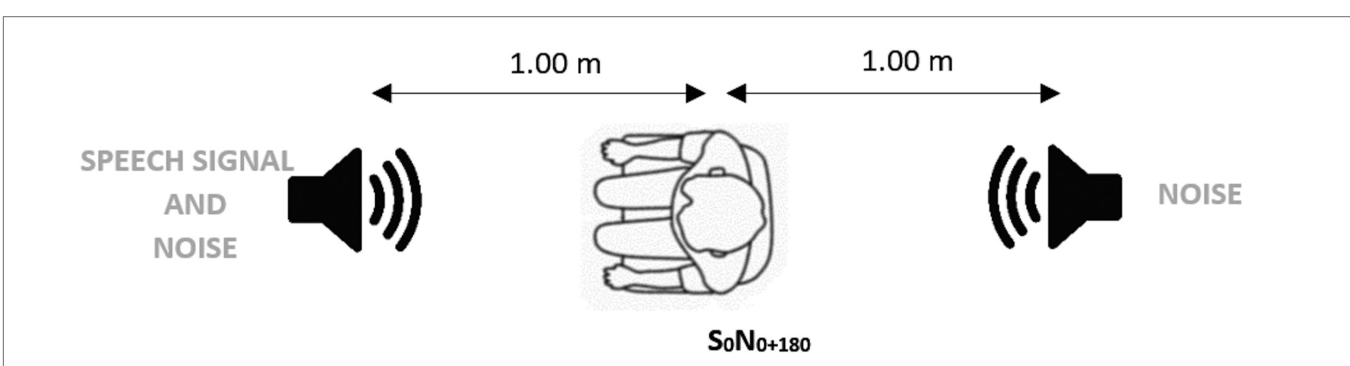

Figure 1. Schematic representation of the setting of audiological testing.

bone conduction hearing threshold was 11.2 dB nHL (range 10.0, 23.3 dB nHL). The median aided air conduction hearing threshold was 25.0 dB nHL (range 18.7, 38.7 dB nHL). Median SDS of the atretic ear 88.6 ± 3.8 dB, with the hearing aid was 52.8 ± 1.9 dB nHL. In the contralateral ear, there was no relevant air-bone gap, and PTAs for air conduction and bone conduction thresholds were within normal limits (PTA ≤ 25 dB). The median ABG significantly improved with Baha 5® Cochlear™, from 51.2 dB nHL (range 40.0, 57.7 dB nHL) to 13.7 dB nHL (range 7.5-23.7 dB), (Wilcoxon Signed Rank test, $p = 0.018$). At SIMT, median SNR without the hearing aid was -5.6 dB (range -7.0, -1.6 dB), while it was -6.4 dB nHL (range -7.8, -2.3 dB nHL) with the hearing aid, which resulted in a statistically significant improvement (Wilcoxon Signed Rank test, $p = 0.027$). Five of seven patients performed the cognitive test with a median score of 37 (range 2-97). Cognitive status did not correlate with either Baha 5® Cochlear™ -aided PTA or Baha 5® Cochlear™-aided ABG (Spearman correlation test, $\rho = 0.2$, $p = 0.741$ and $\rho = 0.1$, $p = 0.873$, respectively).

Discussion

The present study, performed in children with unilateral atresia, highlights the benefits of a bone conduction hearing aid (specifically the Baha 5® Cochlear™) in background noise as measured by the SIMT. The benefit is noticeable in all patients, regardless of cognitive abilities.

In recent studies, chronic conductive unilateral hearing loss has been shown to cause speech intelligibility deficits [24], poor academic performance [17], and overall, a reduced quality of life in the paediatric population [18]. This can be exacerbated in patients with congenital conditions causing hearing impairment, such as aural atresia. These patients must face congenital unilateral hearing loss with crucial impact in the global development of the child. Most probably, patients with unilateral atresia might be "forced" to a unilateral hemispheric dominance if the atretic ear is not stimulated in the first years of life [25]. The consequence can be a reduced benefit perception when a hearing aid is used in adulthood.

In addition, the concomitant presence of poor cognitive performance or attention deficit and hearing loss might have tremendous impact on attention abilities and consequently on scholastic performance of affected children. Moreover, classrooms are known to be extremely noisy environments in which acoustic conditions can be extremely challenging [26].

SIMT is a recently developed hearing test that is useful to evaluate hearing performance in background noise. The test is composed of words familiar to children and consists of an adapted version of the Italian Matrix Test, a test that is becoming more common in clinical and research settings [20,22,27]. In other countries, the simplified test has been shown to be feasible even in the elderly [28].

In the present study, all children were able to perform the test since it is perceived as a game. Obviously, older children were more collaborative and completed the test rapidly. The difference in terms of cognitive abilities did not seem to impair the evaluation nor did it correlate with the hearing results. Indeed, all patients showed better results while using the hearing aid and children with poor cognitive skills seem to have better hearing results. Since the non-aided hearing performance (measured with the PTA) of the present cohort is in line with previous studies [1] and most probably the benefits of a hearing aid can be expected in larger populations of patients with unilateral atresia.

Despite the risk of a possible masking effect of the BCHA on the contralateral normal hearing ear, no patient showed a

Table I. Audiological and cognitive results of the patients.

PT	Age	Sex	Side	AC-PTA AE (dB)	BC-PTA AE (dB)	PTA BCHA (dB)	SDS AE (dB)	SDS BCHA AE (dB)	SRT (dB)	SRT BCHA (dB)	Cognitive score (%)
1	10	F	R	68.75	11.25	18.75	90	60	-5.8	-6.7	37
2	7	F	R	66.25	11.25	25	90	50	-6.9	-6.9	84
3	6	F	R	70	23.3	38.75	90	60	-1.6	-2.3	NA
4	11	M	L	50	10	18.75	90	30	-5.2	-6.1	2
5	10	M	L	58.75	10	21.25	90	40	-7	-7.8	97
6	8	M	R	66.25	11.25	26.25	90	70	-3.8	-6.4	NA
7	11	M	R	62.5	11.25	35	80	60	-5.6	-5.9	14
Mean values	9.00	-	-	63.21	12.61	26.25	88.57	52.85	-5.13	-6.01	46.8
Standard deviation	2.00			6.95	4.74	7.87	3.77	13.8	1.89	1.75	42.08

PT: patient; F: female; M: male; R: right; L: left; AC: air conduction; PTA: pure-tone audiometry; AE: atretic ear; BC: bone conduction; SRT: speech recognition threshold; SDS: speech discrimination score; BCHA: bone conduction hearing aid; NA: not available.

decrease in auditory performance when tested with SIMT. Consequently, the present study consolidates and reinforces the idea that BCHA in unilateral conductive HL might improve speech perception in noise thanks to the summation effect, but the setting of this study prevented investigation of real binaural hearing. In this regard, the evaluation of the possible masking effect of a BCHA on the contralateral hearing should be considered beyond the objectives of the present study, and this effect can deteriorate speech perception due to the reduction of binaural cues.

The main limitation of the study is the small sample size of the cohort considered. Unfortunately, the rarity of the pathology impairs the collection of large cohorts of patients even more when considering the specific target of the study (i.e. patients with unilateral atresia without experience of bone conduction hearing aids). Moreover, the collaboration of children is limited in time thus causing the loss of some data. The objective for the future is to extend the cohort and compare these results with cohorts of patients with longer periods of training or that use BCHA from the first phases of life.

Consequently, the above-mentioned results should be considered preliminary, but they show that a benefit can be perceived by young unilateral atretic patients even immediately after the very first use of the hearing aid. These findings may be even better in patients with longer training periods using the device on daily basis. In addition, the modern bone conduction hearing aids allow extended connectivity via Bluetooth®. This may be of particular interest in the paediatric population since remote communication systems can be connected to multiple devices and teachers can use these features in scholastic environments. In addition, the hearing aid can be connected to smartphones and/or tablets, thus encouraging its use in ludic contexts. In any case, instruments such as the Baha 5® Cochlear™ are useful to increase the signal to noise ratio, suggesting their use in noisy environments. Moreover, this specific device can be worn with a headband, an arch, or implanted with a temporal bone screw or magnetic contacts, allowing children to test the device and its benefits before surgical implantation. The daily experience with these patients is that many children are enthusiastic to at least try a bone conduction hearing aid. Nonetheless, it is common experience that aesthetic reasons might play a role in reducing the use of the hearing aid and drive patients to become non-users.

These preliminary findings point towards the proposal of early rehabilitation of the unilateral atresia using bone conduction hearing aids. The benefit may be extremely important in a scholastic environment to improve the signal to noise ratio and consequently to support learning abilities.

Conclusions

The present study, conducted on a relatively small cohort of patients affected by unilateral aural atresia, shows that SIMT can be considered a feasible instrument to evaluate hearing performance in noisy context. Baha 5® Cochlear™ was shown to be a valuable device that can provide benefit in complex hearing conditions, and this benefit can be even more relevant for children with poor cognitive abilities. These findings suggest that early rehabilitation with a bone conduction hearing aid should be considered and discussed with patients' families as promising results have been documented. In conclusion, it is likely time to address the problem of good hearing in patients with unilateral atresia from another side, the affected one.

Conflict of interest statement
The authors declare no conflict of interest.

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Author contributions
DB, FS, PT: conceived the idea of the study; DB, FS, FM, SM, AA: developed the theory and performed the tests; DB, FS, DC, EZ, AM, RB, PT: verified the analytical methods and supervised the findings of this work. All authors discussed the results and contributed to the final manuscript.

Ethical consideration
This study was approved by the Institutional Ethics Committee of the Azienda Ospedale Università Padova (protocol number 1077).

The research was conducted ethically, with all study procedures being performed in accordance with the requirements of the World Medical Association's Declaration of Helsinki.

All patients or parents of children gave written informed consent for study participation and data publication.

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