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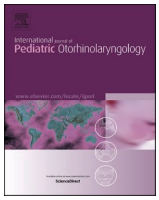

Patient reported outcome measures in unilateral aural atresia treated using a transcutaneous bone conduction implant (The Cochlear Baha Attract®)

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ABSTRACT

Objectives: Unilateral aural atresia (UAA) is a congenital condition that is associated with maximal conductive hearing loss. The primary objective of this study was to assess the long-term compliance, complications, and quality of life of pediatric patients with UAA who had transcutaneous bone conduction hearing implants (TCBI).
Methods: Retrospective consecutive case series at a single centre between 2014 and 2018. Inclusion criteria consisted of UAA patients between 5 to 17 years-old receiving the Cochlear Baha Attract ® device. Demographic and audiologic data was extracted from charts. A prospective telephone survey was done, with patients and their families completing the Glasgow Children's Benefit Inventory (GCBI). Basic descriptive statistics, paired t-tests, and a univariate analysis were completed.
Results: Data was successfully collected from all 9 eligible children who received the Cochlear Baha Attract ® device for UAA (100%). The mean follow-up duration was 33 months after TBCI (9–60 months). The mean daily use was 7.7 h/day. Pure tone average and mean speech in noise scores were both significantly improved when comparing the unaided condition to the aided condition with TCBI ($p < 0.001$). The majority (89%) of patients had an improvement in GCBI; the median GCBI score was +14.6, indicating overall positive benefit. A linear regression showed no demographic variables were significant for mean daily use or GCBI scores.
Conclusion: This preliminary study showed that patients with a TCBI for UAA had high long-term compliance and daily usage rates. TCBI improved the quality of life for the majority of patients and significantly improved hearing measures.

1. Introduction

Aural atresia is a congenital malformation of the external auditory canal often associated with malformation of the middle ear. The incidence is reported at 1 in 10,000 to 20, 000 live births [1]. While aural atresia can occur in isolation, it is often associated with various syndromes.

Patients with unilateral aural atresia (UAA) can have maximum conductive hearing loss on the affected side. Compared to hearing peers, patients with unilateral hearing loss are at increased risk for educational and behavioral difficulties [2]. Specifically, those with UAA have higher chances of speech and learning difficulties [3,4].

Early use of bone conduction hearing aids on soft-bands for binaural hearing are therefore recommended for patients with UAA. For later surgical interventions, osseo-integrated devices have overtaken canalplasty as first-line treatment. Nadaraja et al.'s systematic review found patients with bone-anchored hearing devices (BAHD) had better audiologic outcomes and less surgical complications than canalplasties [5]. Transcutaneous bone conduction implants (TCBI), where an implanted magnet is paired with an external processor, allow for good aesthetic outcomes with less skin breakdown than traditional percutaneous implants [6]. Furthermore, TCBI has comparable hearing outcomes to earlier generations of BAHDs [7].

The primary objective of this study was to review and characterize compliance and quality of life in patients with UAA who underwent TCBI using a specific device, the Cochlear Baha Attract ®. Secondary objectives included examining audiologic outcomes and potential associations between demographic variables with increased hearing aid use or quality of life.

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2. Materials and methods

This cross-sectional observational study was approved by our institution's Clinical Review Ethics Boards *(No:* H19-02115) Informed written consent was obtained from parents or guardians, and assent from children above the age of 7.

2.1. Subjects

Patients were recruited from a single tertiary academic centre. Inclusion criteria consisted of: patients between 5 to 17 years-old with UAA and had TCBI surgery with a unilateral Cochlear Baha Attract ® device at least 4 months prior to recruitment. Patients and their families were contacted by telephone to participate in a survey and to complete the Glasgow Children's Benefit Inventory (GCBI).

The GCBI is a patient reported quality of life questionnaire designed to be used post-intervention consists of 24 questions answered using a five-point Likert scale. The responses were scaled and averaged to give a score with a range of −100 (poorest outcome) through to +100 (best outcome) [8]. The GCBI was then also scored on four subscales: "Psycho-social," "Physical health," "Behavior," and "Vitality" [9].

2.2. Data collection and analysis

Demographic factors, clinical history, details of the surgery, and baseline as well as the most recent audiograms were extracted from those that decided to participate in the telephone survey. All hearing assessments were completed by registered audiologists at the same tertiary academic centre, with baseline unaided ears pre-surgery compared to recent hearing assessments of patients' aided ears with BAHDs. All hearing assessments were done at 500 kHz, 1000 kHz, 2000 kHz, and 4000 kHz. Speech in noise testing was completed with Pediatric AzBio sentences presented using recorded voice, but testing may have been modified depending on patients' development. The telephone survey included assessing the amount of daily self-reported TCBI use, level of satisfaction with device, and complications or drawbacks. Basic descriptive statistics were used for demographic factors. Paired t-tests and a univariate linear regression were analyzed with R (Version 5.3).

3. Results

In total, 18 patients were identified at our centre that received bone conduction hearing implants over the study period; 15 of these were for UAA. A total of nine patients had received the Cochlear BAHA Attract ® device for UAA and were considered eligible. 100% (9/9) of these subjects consented to participate in the study and were included (median age 8, 44.4% males). The attrition and response rate of patients are shown in Fig. 1. The mean follow-up duration after transcutaneous implant surgery was 32.8 months (9–60 months). The mean self-reported daily use of their TCBI was 7.7 h/day. Patients demographic and clinical features are summarized in Table 1.

Patients who had full audiometry testing showed an improvement in their aided pure tone averages (PTA) across 500–4000 Hz compared to their unaided baseline, as well as with mean speech in noise scores ($p < 0.001$). The majority of patients (87.5%) had a post-operative aided hearing level suggesting mild hearing loss (15–40 dB), with the remaining patient having aided hearing within the normal range post-operatively (<15 dB). One patient who underwent the surgery was unable to have a complete hearing assessment, but was included in the analysis for their demographic and GCBI results.

For the patient-reported GCBI, the majority of patients had an overall quality of life benefit (89%). The median GCBI across all domains for patients was +14.6 ([6.2, 27.1] 25th, 75th percentile). Fig. 2 displays the GCBI scores for each individual subscale. Two specific questions were scored and averaged: "Has your child's operation affected his/her progress and development" and "Has your child's operation affected his/her learning". A median score of 75 ([50, 100] 25th, 75th percentile) for the former and 55.6 ([50, 75] 25th, 75th percentile) for the latter were obtained.

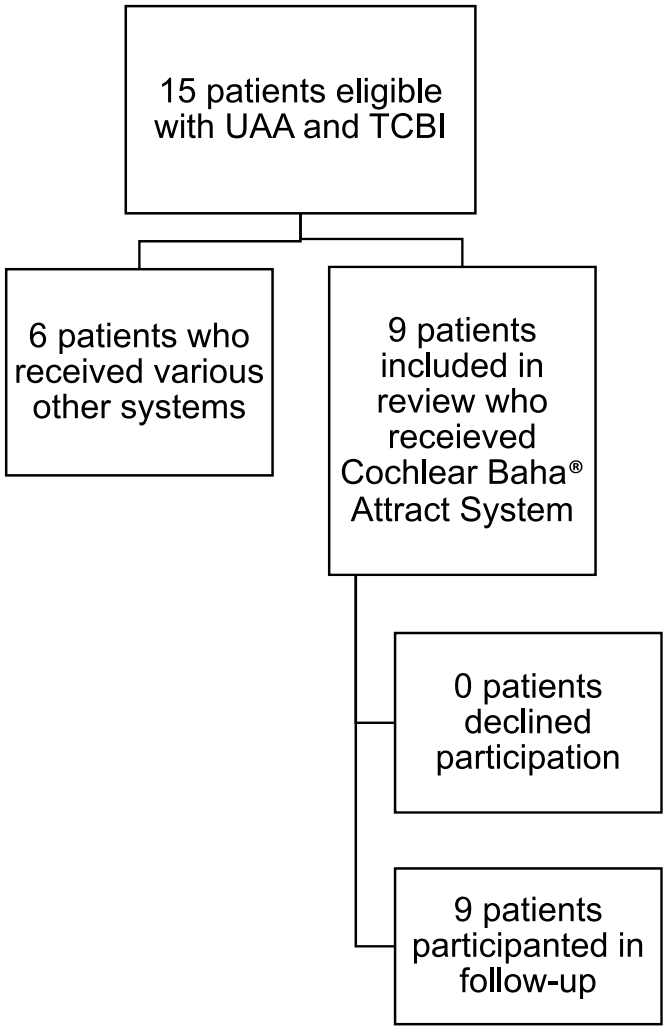

Fig. 1. Attrition and response rate of patients who underwent a transcutaneous bone conduction implant (TCBI) for unilateral aural atresia (UAA) from June 2014–December 2018 and were contacted to complete the surveys in July of 2019.

One patient required revision surgery for poor wound healing. The complications reported by three patients' families included: discomfort with prolonged use, headaches, and that the device's battery closure easily broke.

When asked about their overall satisfaction as a parent with respect to the TCBI experience all caregivers stated "satisfied" or "very satisfied", except for one. When asked if they would recommend surgery to other parents with the same condition, all caregivers stated "recommend" or "strongly recommend", except for one.

Linear regression of the following demographic factors showed no variables significantly associated with mean daily use or GCBI score: age at time of surgery, gender, side of TCBI, follow-up period length, or average change in hearing over 500 kHz, 1000 kHz, 2000 kHz and 4000 kHz.

4. Discussion

This is the first study to look at patient-reported benefit after TCBI for pediatric patients with unilateral aural atresia. Overall, our patient population reported benefit with TCBI and had an average of nearly 8 h

Table 1
Clinical demographics of patients who underwent transcutaneous bone conduction hearing implants for unilateral aural atresia. Average Change in Hearing over 500 kHz, 1000 kHz, 2000 kHz, and 4000 kHz was defined as baseline unaided audiograms of patients compared to aided audiograms after bone anchored hearing device.

Patient number	Age at time of surgery	Gender	Comorbidities	Side of TCBI	Follow-up period (months)	Average Change in Hearing over 500 kHz, 1000 kHz, 2000 kHz, and 4000 kHz (dB)	GCBI	Complications reported
1	11	Male	CHARGE syndrome Goldenhar syndrome	Left	8	N/A	20.8	
2	11	Female		Left	24	42.5	−27	Discomfort with prolonged use
3	16	Female	Hemifacial microsomia	Left	12	41.3	4.1	
4	17	Female		Right	26	33.8	22.9	
5	5	Female		Right	59	43.8	8.33	
6	8	Female	Goldenhar syndrome	Left	36	38.8	18.75	
7	7	Male		Right	30	37.5	31.2	Headaches
8	8	Male		Left	42	42.5	35.4	
9	7	Male		Right	58	38.8	16.6	Device easily breaks with use

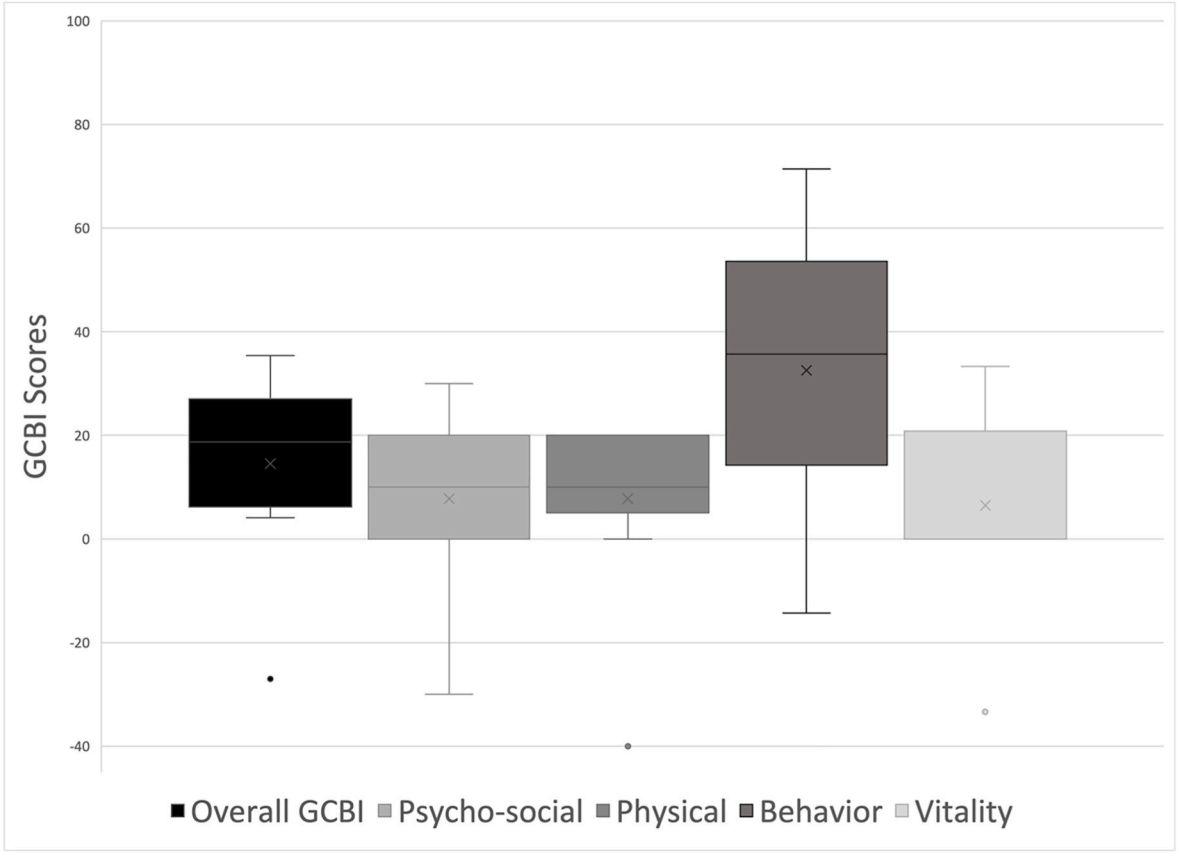

Fig. 2. Box and whisker plot depicting the overall GCBI scores of all subjects and the scores for each of the four domains: "Psycho-social," "Physical health," "Behavior," and "Vitality. The boxes define the 25th and 75th percentiles, the whiskers define the 10th and 90th percentiles, and the dots define the 5th and 95th percentiles.

of self-reported daily use at various time-frames after surgery. Only patients who had the Cochlear Baha ® Attract System anchored were included to remove the potential confounder of various BAHDs.

Our results showed significant improvement in average PTA and mean speech-in-noise after TCBI (Table 1). This was as consistent with other similar reports in the literature; Lippmann et al. found that of their 41 implantations for pediatric aural atresia, mean PTAs improved from 63.7 to 9.6 [10]. This is comparable to our mean PTA change unaided vs aided ears over 500, 1000, 2000, and 4000 Hz of 39.9. A systematic review of BAHDs for congenital hearing loss found consistent gain in audiologic speech reception thresholds and speech discrimination [11].

The majority of our patients had reported benefit with TCBIs for UAA (Fig. 2). The largest improvement was in the behaviour subscale, with specific questions assessing "progress and development" "and "learning" showing maximum benefit. Many of our families also reported most frequent use when the patients were at school.

While less profound than in children with unilateral sensorineural hearing loss (SNHL), children with UAA and its associated conductive

hearing loss are academically impacted [4]. Social challenges and emotional implications have not been as well studied. Our reported complications included discomfort with prolonged use and headaches, but no patients reported social anxieties with TCBI use. The GCBI psycho-social subscale was also similar to our physical and vitality subscale.

Doshi et al. examined quality of life after BAHDs in eight pediatrics patient with single-sided sensorineural deafness. Similarly to this study, they found an overall GCBI benefit, although their median score of +47.5 was higher than what was reported in this study [12]. In an adult population of patients with BAHDs, Martin et al. reported a median score of +11 which is similar to our results [13]. A study that examined 105 patients with TCBIs found that of the six children who answered the Speech, Spatial, and Qualities of Hearing Scale, there was a 22% improvement in mean scores after aiding [14]. Other pediatric otolaryngologic interventions have shown median GCBi scores ranging from +19.5 for submucosal diathery of inferior turbinates [15] to +35.1 for septoplasty [16].

There is a paucity of literature in regards to TCBI in the pediatric population. The authors would support the use of TCBI in patients with UAA based on the results of this study, as patients reported positive benefits at various time frames and high usage. It is reassuring that, beyond one patient, most families reported they would recommend and were satisfied with their TCBIs.

Although TCBIs have gained in popularity, more work could be done to improve the shared-decision making process with these patients and their families. Benefits of shared-decision making include reduced decisional conflict, realistic expectations of outcomes, and improved feelings of support [17]. In particular, caretakers may have decisional conflict regarding the consideration of surgical hearing aids for pediatric patients. A study examining twenty-three caregivers found that 43.5% reported significant decisional conflict when considering BAHD for UAA [18]. This current study helps examine patient perspectives and may help providers with the informed consent process.

Limitations of this study include that this was a single-centre study with patients recruited at a quaternary centre. The results may not be applicable elsewhere. Our retrospective analysis and self-reported survey administration may be vulnerable to bias inherent within this study design, including self-selection and recall bias. Furthermore, the authors acknowledge that a larger sample size would provide more robust data and conclusions, although there were was fortunately a 100% participation rate of the eligible subjects in this consecutive case series. The design of this study to include only a single specific device fulfilled the need for intervention consistency, but this design trades that strength off against the potential weakness of reduced generalizability of the results to other TCBI devices for the same indication. Our follow-up periods could have been more substantial to gain true long-term outcomes. Finally, more significant results could have been obtained if patient-reported GCBI scores from a control group of patients who elected not to have surgery and continued soft-band use were included.

5. Conclusions

This is the first study to examine patient-reported outcome measures after TCBI for UAA in a pediatric population. The results demonstrated these patients had excellent improvements in audiological outcomes and overall quality of life benefit. TCBIs usage compliance was high at a long follow-up duration from their procedure and these devices warrant continued consideration as a treatment option for this population.

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Declaration of competing interest

None.

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