Early Outcomes of a New Active Transcutaneous Bone Conduction Implant in Children with Microtia-Atresia

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[Redaction]

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Statement of Originality

This is to certify that the content of this thesis is my own work and has not been submitted, in whole or in part, for any other degree or purpose.

I certify that the intellectual content presented in this thesis is the product of my own work. All sources of information and assistance received during the preparation of this thesis have been appropriately acknowledged.

Dr Naomi (Natasha) Niles

29 September 2025

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To my family and loved ones - thank you for your constant encouragement, patience and understanding. Your belief in me has been my anchor through every challenge and triumph.

And finally, to my beloved niece Shemara, who inspires every page of this work, and her parents, whose strength, love, and unwavering dedication light her path - this is for you. May it help, in some small way, to build a world where every child with microtia can reach their full potential.

Authorship Attribution Statement

Chapters 2 and 3 of this thesis have been formatted for submission to the *International Journal of Pediatric Otolaryngology* and the *Australian Journal of Otolaryngology* respectively for publication. I designed the aforementioned studies with my supervisors, collected and analysed the data and prepared the manuscripts. Furthermore, I understand that, if this thesis is successful, it will be lodged with the University Librarian and made available for immediate public access.

Dr Naomi (Natasha) Niles
26 September 2025

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As supervisors for the candidature upon which this thesis is based, we hereby certify that the authorship attribution statement provided above is accurate. We further confirm that this thesis is of an examinable standard and does not exceed the prescribed word limit.

Professor Daniel Steffens
26 September 2025

Associate Professor Payal Mukherjee
26 September 2025

Artificial Intelligence Statement

During the preparation of this thesis, the author used CoPilot (Microsoft 2025, https://copilot.microsoft.com) and ChatGPT-5 (OpenAI 2025, https://chatgpt.com) to assist with refining text, including enhancing clarity and improving sentence structure. The author confirms that where the text was modified by generative AI, the author reviewed the content for possible errors, inaccuracies, and biases. The author takes full responsibility for the submitted thesis, confirms the work is their own and has used generative AI within the parameters of use, as set out in the University of Sydney Generative AI Guide for Researchers.

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Glossary

ABG	Air-Bone Gap
AC	Air Conduction
atBCI	Active Transcutaneous Bone Conduction Implant
BC	Bone Conduction
BCD	Bone Conduction Devices
BC-FMT	Bone Conduction Floating Mass Transducer
BI300	Cochlear™ osseointegrated implant screw used for Baha/Osia fixation
BKB	Bamford-Kowal-Bench (sentence test)
CHL	Conductive Hearing Loss
CMHL	Conductive or Mixed Hearing Loss
CT PTB	Computed Tomography of the Petrous Temporal Bones
dB	Decibels
dB HL	Decibels Hearing Level
EAC	External Auditory Canal
FDA	U.S. Food and Drug Administration
FG	Functional Gain
HEAR-QL	Hearing Environments and Reflection on Quality-of-Life (questionnaire)
kHz	Kilohertz
MRI	Magnetic Resonance Imaging
PEACH	Parents' Evaluation of Aural/Oral Performance in Children
PROMs	Patient-Reported Outcome Measures
PTA	Pure Tone Average
SIN	Speech-in-noise
SNHL	Sensorineural Hearing Loss
SNR	Signal-to-Noise Ratio
SPINE	Speech Perception in Noise (sentence test)
SRT	Speech Reception Threshold
SSD	Single-Sided Deafness
SSQ	Speech, Spatial and Qualities of Hearing Scale
TGA	Australian Therapeutic Goods Administration
TPF	Temporoparietal Fascia
uSNHL	Unilateral Sensorineural Hearing Loss

Thesis Abstract

Background

Microtia-atresia encompasses a spectrum of congenital malformations affecting the auricle, external auditory canal, and, in some cases, the middle ear, leading to conductive hearing loss. Even in unilateral cases, which are more common, microtia-atresia can adversely affect speech, language, academic performance, and quality of life. Bone conduction hearing devices remain the mainstay of hearing habilitation. Active transcutaneous bone conduction implants (atBCIs) offer advantages in comfort, fewer skin complications, reduced feedback, and better high-frequency transmission than traditional passive devices (1). The Osia® (Cochlear™ BAS, Gothenburg, Sweden) is a relatively recent atBCI, with regulatory approval for implantation in children aged 5-12 years only granted in Australia and the United States in 2024. Evidence in this younger population remains sparse, and no systematic reviews or Australian outcome data have been published to date.

Aims and Purpose

The overarching purpose of this thesis is to evaluate the role of active transcutaneous bone conduction implants in children with microtia-atresia, with a focus on the Cochlear Osia.

The specific aims of this thesis are to:

1. Systematically review the published evidence on paediatric Osia outcomes, focusing on safety, audiological benefit, and patient (or parent)-reported outcome measures (PROMs).
2. Present the first Australian case series of children 12 years of age or younger with microtia-atresia undergoing Osia implantation, evaluating safety, audiological benefit, and functional listening outcomes.

Methodology

- **Chapter 1** outlines the clinical context and significance of microtia-atresia, including anatomy, embryology, physiology, functional impact, and current management, to contextualise the subsequent chapters.
- **Chapter 2** presents a systematic review of paediatric Osia outcomes, focusing on complications, audiological outcomes and PROMs.
- **Chapter 3** reports a retrospective case series of initial Australian experience with Osia implantation in children with microtia-atresia, most of whom underwent concurrent single-stage alloplastic auricular reconstruction.
- **Chapter 4** synthesises findings and discusses clinical implications and research priorities.

Results

- **Chapter 2 (Systematic Review):** Paediatric Osia implantation is associated with a favourable safety profile and clear audiological benefit. However, existing evidence is limited to small, heterogeneous observational studies with variable follow-up and a paucity of functional outcome data, underscoring the need for multicentre prospective research with standardised outcome measures.
- **Chapter 3 (Case Series):** Osia implantation is feasible and safe in carefully selected children with microtia-atresia. Implantation produced significant improvements in aided thresholds, with functional gains comparable to published adult and paediatric data. Post-operative Parents' Evaluation of Aural/Oral Performance in Children (PEACH) scores indicated enhanced listening performance in both quiet and noise. Parents consistently highlighted improved communication, reduced listening fatigue, greater classroom participation, and higher compliance compared to pre-operative devices.

Conclusion

This thesis supports Osia as a safe and effective option for hearing habilitation in children with microtia-atresia. Early implantation may help mitigate developmental and educational risks associated with prolonged unilateral conductive hearing loss. Although constrained by small cohort sizes – reflecting both the rarity of microtia-atresia and the recent approval of Osia in younger children – this thesis provides the first synthesis of international paediatric Osia outcomes and reports the first Australian case series of its kind. As such, the findings contribute uniquely to the literature, offering early evidence of feasibility, safety and functional benefit in this population. They also highlight the need for larger prospective studies with standardised outcome measures and long-term follow-up to validate these results and inform best practice in paediatric microtia-atresia management.

Chapter 1: Introduction

Background and Context

Congenital aural atresia is one of the most common congenital anomalies of the head and neck region. It is characterised by narrowing or absence of the external auditory canal (EAC), usually accompanied by abnormalities of the middle ear (Figure 1). The condition is often associated with malformations of the pinna (microtia), which can range from mild hypoplasia to complete absence (anotia) (Figure 2). Due to their frequent co-occurrence, the term *microtia-atresia* is commonly used. The global prevalence of microtia-atresia varies from 0.8 to 17 per 10,000 births, with higher rates among Hispanics, Asians, Pacific Islanders, Native Americans and Andeans. The condition is unilateral in most cases (77-93%); more often affecting the right ear; and has a slight male predominance. Between 20% and 60% of children with microtia-atresia have associated anomalies or an underlying syndrome, such as hemifacial microsomia, Goldenhar syndrome, Treacher Collins syndrome and Trisomy 21 (2-4).

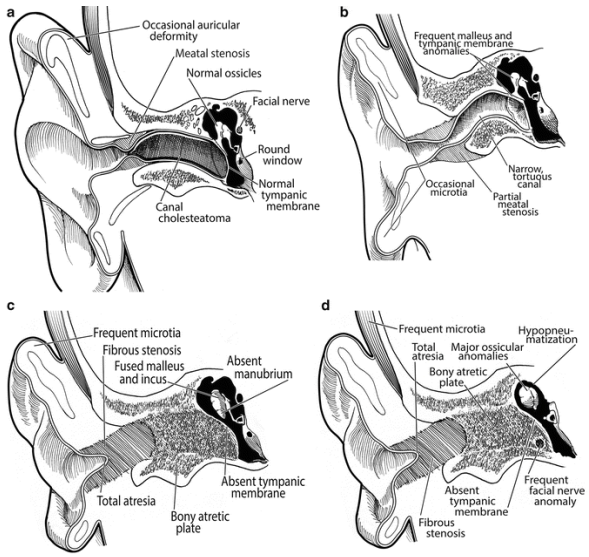


FIGURE 1. Types of Congenital Aural Atresia (based on the Schuknecht classification system). **A)** Type A: Meatal atresia. **B)** Type B: Partial atresia. **C)** Type C: Total atresia. **D)** Type D: Hypopneumatic total atresia; *from Shoman et al 2014* (5).

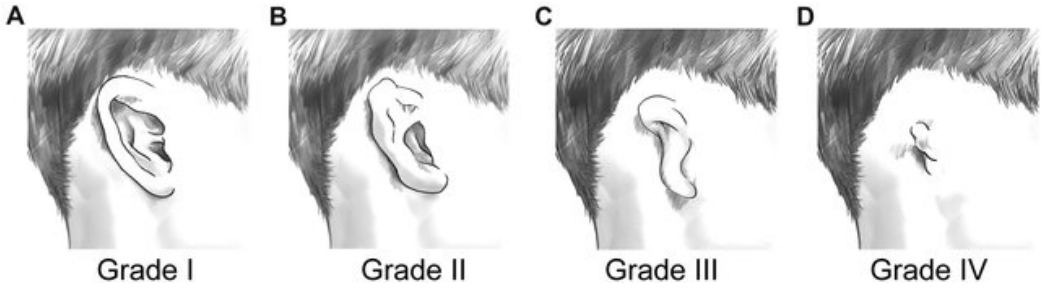


FIGURE 2. Microtia Grading. **A)** Grade I. All the external ear structures are identifiable, except for a smaller but present auricle and external ear canal. **B)** Grade II. A partially developed ear with stenotic external ear canal. **C)** Grade III. Absence of external ear with only a peanut-shell structure remaining. **D)** Grade IV. No external ear or ear canal; *from Cheng et al 2021* (6)

Beyond the structural malformation, microtia-atresia carries substantial psychosocial and developmental consequences. Children often experience stigma related to the visible anomaly, and many face the emotional and physical burden of undergoing multiple reconstructive surgeries throughout childhood (2). Furthermore, the associated conductive hearing loss (CHL) can significantly impact speech, language, learning and social participation in children, underscoring the importance of early and effective intervention (7).

Anatomy and Embryology

The ear is anatomically and functionally divided into three distinct regions: the external, middle, and inner ear (Figure 3). The *external ear* consists of the cartilaginous pinna (also referred to as the auricle) and external auditory canal (EAC). Together, these structures funnel sound waves toward the tympanic membrane, which vibrates in response. The *middle ear* is an air-filled cavity that houses the ossicles, three small bones that transmit vibrations from the tympanic membrane through the oval window. The *inner ear* comprises the cochlea and the vestibular system. Vibration of the oval window generates movement in the endolymph of the cochlea, stimulating the sensory hair cells of the organ of Corti and transducing mechanical energy into electrical signals that are transmitted to the auditory cortex via the cochlear nerve (8).

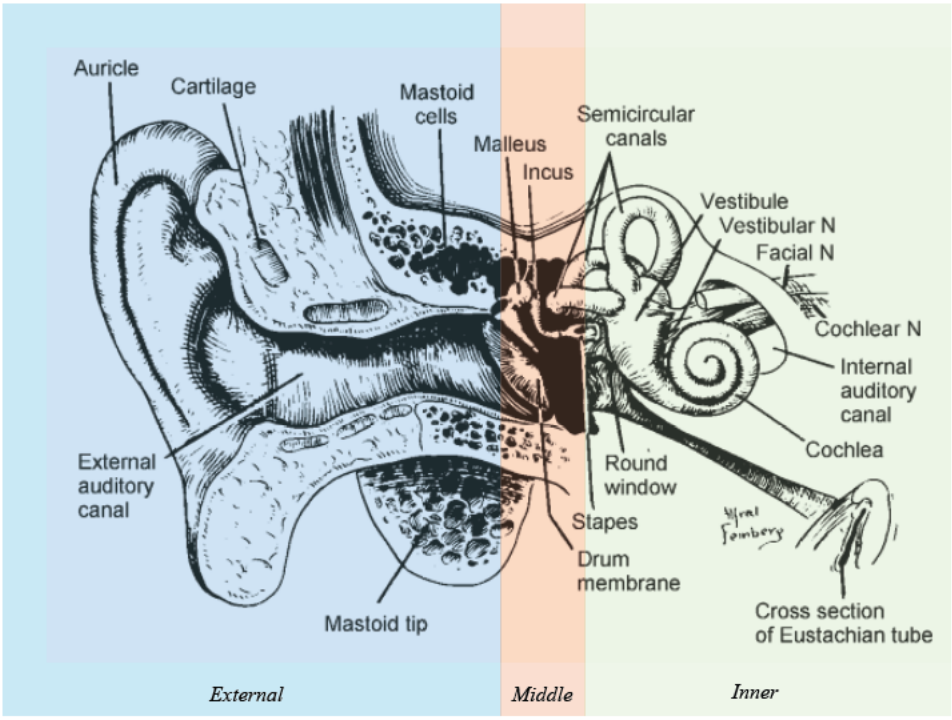


FIGURE 3. Anatomy of the Ear; *from Davis & Silverman 1970* (9)

During embryogenesis, a series of paired swellings, known as the branchial arches, form along the ventrolateral surface of the embryo and give rise to many of the head and neck structures. The external and middle ear share a common embryological origin in the first and second branchial arches, explaining the frequent association of microtia and aural atresia (Figures 4 and 5) (2). In contrast, the inner ear develops independently from the otic placode and is typically spared, accounting for the predominantly conductive nature of hearing loss seen in these children. However, inner ear abnormalities are reported in 11-30% of cases, particularly in those with underlying syndromes, resulting in mixed hearing loss (10).

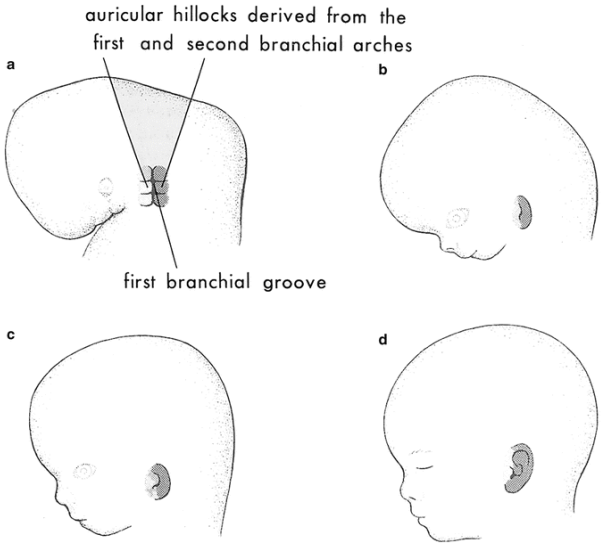


FIGURE 4. Development of the Auricle; *from Moore & Persaud 1993* (11).

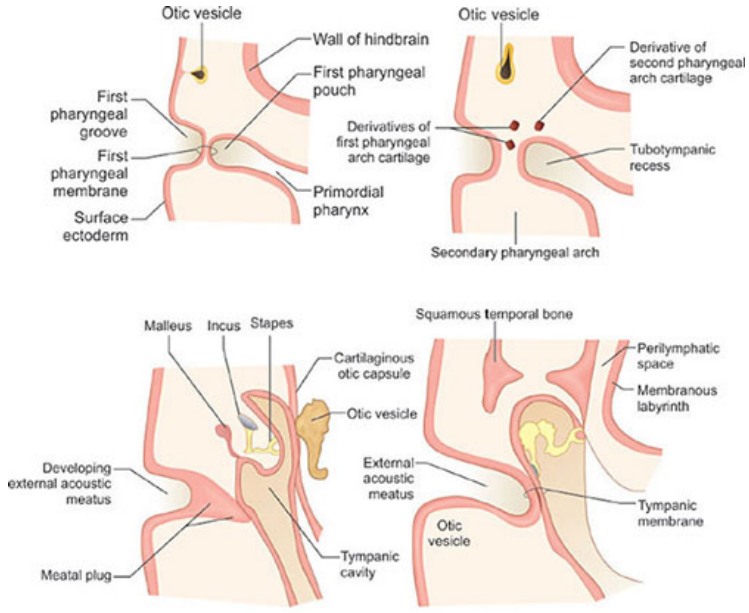


FIGURE 5. Development of the external auditory canal, middle and inner ear; *from Bhalodiya 2025* (12).

Hearing Physiology and Hearing Loss

Hearing loss may be conductive, sensorineural, or mixed. *Conductive hearing loss* (CHL) results from impaired sound transmission through the EAC or middle ear due to structural abnormalities or blockages. In aural atresia, where the canal and middle ear structures are malformed, conventional air conduction aids are generally ineffective. Thus, bone conduction

devices, which directly stimulate the cochlea via skull vibrations, are the mainstay of rehabilitation.

Sensorineural hearing loss (SNHL) results from pathology of the cochlea or auditory nerve pathways, commonly associated with genetic conditions, aging, noise exposure and infections. Cochlear implants may be used in moderate-to-profound cases, converting sound to electrical impulses that directly stimulate the auditory nerve. Single-sided deafness (SSD) refers to a pattern of profound unilateral SNHL with normal contralateral hearing. In these cases, BCDs can be used to transmit sound from the deaf side to the functional cochlea as vibrations of the skull stimulate both cochleae.

Mixed hearing loss (MHL) refers to concurrent conductive and sensorineural hearing loss in the same ear, with rehabilitation tailored to the individual audiometric profile.

Neuroplasticity and Auditory Deprivation

The clinical impact of hearing loss depends not only on its type and severity, but also on the developing brain's capacity to adapt. Hearing relies on the coordinated activity of multiple brain regions, including the primary auditory cortex within the anterior temporal lobe, the prefrontal cortex, and the inferior parietal cortex, together with extensive cortical and subcortical networks (13). The adaptability of these systems is underpinned by neuroplasticity - the brain's ability to reorganise its structure and function in response to internal and external stimuli. This capacity peaks between two and four years of age, marking a 'sensitive period' for auditory development (14, 15). Large-scale studies demonstrate that consistent, balanced auditory input from both ears during this period is critical for establishing the neural architecture underlying higher-order auditory processing (16-21). This

sensitive window provides a strong impetus for early detection and intervention in children with hearing loss, as delays in auditory access may result in long-lasting deficits that are only partially reversible once cortical maturation has occurred.

Historically, unilateral hearing loss (UHL) - the most common pattern in children with microtia-atresia - was considered benign and not in need of intervention. However, since the late 1980s a growing body of evidence has challenged this view, demonstrating that children with UHL are at risk of delayed expressive and receptive language development, reduced vocabulary, and poorer academic outcomes than their normal hearing peers (22-30).

The disadvantages of UHL are best understood by contrast with the benefits of binaural hearing. Hearing with two ears is not redundant; rather it provides specialised mechanisms that enhance auditory processing. These include the *head shadow effect*, which improves the signal-to-noise ratio by favouring the ear with the clearer input; *binaural summation*, which enhances loudness perception and speech detection thresholds; *binaural squelch,* which enables the central auditory system to separate speech from competing noise using interaural timing and intensity cues; and *sound localisation*, which relies on subtle interaural differences to determine sound origin. Collectively, these mechanisms underpin effective communication in noisy and complex listening environments such as classrooms and playgrounds, where children spend much of their time. Their absence leads to difficulties with speech understanding in noise, impaired localisation, increased listening effort and fatigue, and even safety risks (31).

Functional Outcomes of Children with Microtia-Atresia

Several retrospective studies highlight the real-world consequences of unaided microtia-atresia. Smit et al. found that 28% of children with unilateral microtia-atresia required additional classroom support, with over 30% repeating a grade, predominantly during early

primary school years (3). Similarly, Jensen et al. found that almost a third of children with unilateral microtia-atresia experienced learning difficulties, compared to none in their bilateral SNHL cohort who had been more consistently fitted with amplification (32). Kesser et al. observed that nearly two-thirds of children with microtia-atresia required classroom support, including wireless microphone systems, speech therapy, special education or individualised learning plans (33). Speech and language delays are also common, with over 40% of children requiring intervention (3, 32). More broadly, hearing loss is linked with poorer health-related quality of life and psychosocial wellbeing (34-36). These findings emphasise the importance of timely and effective hearing habilitation.

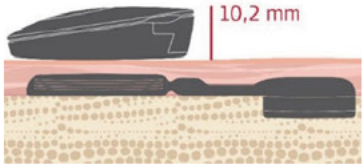
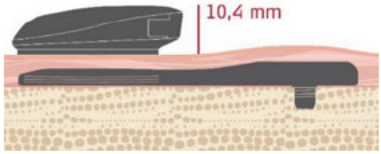
In addition to auditory deprivation, broader child-level factors may also influence functional outcomes in microtia-atresia. Neurodevelopmental differences, cognitive profiles, and co-occurring syndromic diagnoses commonly seen in microtia-atresia, such as Treacher Collins syndrome, Goldenhar syndrome, or Trisomy 21, may compound listening, learning, and communication challenges independent of hearing status. These comorbidities can affect speech and language acquisition, auditory attention, behavioural regulation, and engagement with rehabilitation. Although inconsistently reported across studies, these factors reinforce the importance of multidisciplinary assessment and tailored management alongside timely hearing habilitation.

Management of Hearing Impairment in Microtia-Atresia

The management of children with microtia-atresia has two parallel goals: hearing habilitation and reconstruction of the auricle. Options for hearing habilitation include atresiaplasty, non-implantable bone-conduction devices (BCDs) and surgically implanted BCDs (Table 1). Atresiaplasty (the surgical creation of a patent EAC) may improve hearing in selected patients, but its success is highly dependent on careful patient selection - specifically, the

presence of normal inner ear function and adequately developed middle ear anatomy. Moreover, the procedure carries notable risks, including facial nerve injury, canal restenosis, chronic infection, and often suboptimal auditory outcomes (37-39). Consequently, many patients continue to require hearing amplification even after surgery. Given these limitations, BCDs remain a cornerstone of management in this population (40).

TABLE 1. Types of Bone Conduction Devices

Non-implantable BCDs	Surgically implanted BCDs	
	Passive	**Active**
		
Bone conduction sound processor worn on a softband (Cochlear Baha or Oticon Ponto sound processors) (41).	Passive transcutaneous bone conduction implant (Baha Attract) (42).	Active transcutaneous implant with a floating mass transducer (MED-EL BONEBRIDGE [pictured], Oticon Sentio) (42).
		
Adhesive gel pad retained BCD (MED-EL ADHEAR) (43).	Passive percutaneous bone conduction implant (Baha connect [pictured], Oticon Ponto) (42).	Active transcutaneous implant with a piezoelectric transducer (Cochlear Osia) (42).

BCD = Bone Conduction Devices

Non-implantable systems include those worn on a headband (e.g. Cochlear Baha or Oticon Ponto), via an adhesive gel pad (e.g. MED-EL ADHEAR), or as spectacle-mounted or hat-mounted devices. These options provide early access to sound for young children and can provide valuable insight into the anticipated benefits of implanted devices.

Surgically implanted BCDs can be categorised into passive and active systems (Table 1). Traditional passive systems rely on an *externally* worn processor-transducer that converts sound into vibrations, which are then transmitted to the skull via either a percutaneous (skin-penetrating) abutment or a transcutaneous (magnet-based) connection. While effective, passive systems have several limitations. These include increased device feedback, due to the close proximity of the external processor and transducer; wound complications with percutaneous systems, related to the skin-penetrating abutment; and reduced sound transmission with transcutaneous systems, caused by energy loss through the intervening soft tissue (44).

Active bone conduction hearing implants (atBCIs) represent a significant advancement in auditory rehabilitation. These systems consist of an externally worn processor, retained by a subcutaneous magnet, which captures and digitally processes sound. The processed signal is transmitted transcutaneously to an *internal* (subcutaneous) implant and transducer, which directly vibrates the skull (Figure 6). By eliminating skin-penetrating abutments, atBCIs reduce soft tissue complications, while direct bone coupling bypasses the attenuating effects of skin and subcutaneous tissue, resulting in improved sound transmission, particularly at high frequencies.

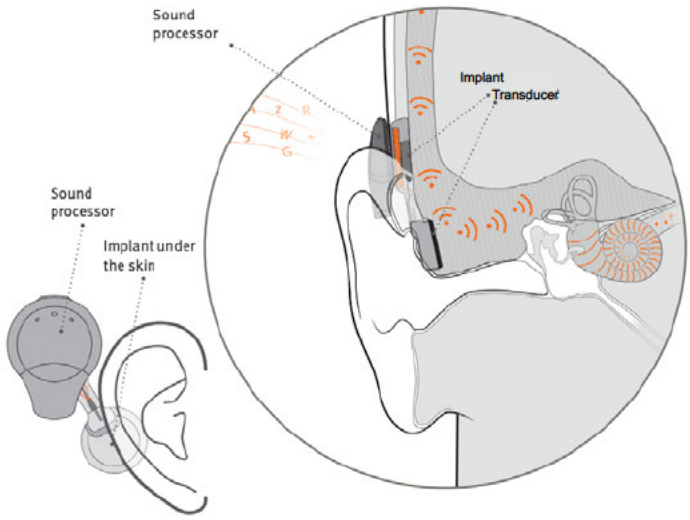


FIGURE 6. Active Transcutaneous Bone Conduction Implant, demonstrating externally worn processor connected to a subcutaneous implant and transducer that sends vibrations directly to the skull and cochlea; *adapted from Oticon Medical 2025* (45).

The BONEBRIDGE® (MED-EL®, Innsbruck, Austria) was the first atBCI to enter the market, released in 2012 as the BCI 601. For several years it remained the only device of its kind until the introduction of the Osia® (Cochlear™ BAS, Gothenburg, Sweden) in 2019 (46). Its transducer required drilling of an 8.7 mm bony recess for fixation. In children, this posed technical challenges due to thinner skull bones (approximately 3.5 mm in the retrosigmoid region of a three-year-old), raising concerns about sigmoid sinus injury, dural exposure, cerebrospinal fluid leak, and further intracranial sequelae (47, 48). Addressing these limitations, MED-EL released a second-generation implant in 2019, the BCI 602, with a reduced transducer profile and design modifications to improve safety and ease of implantation in both paediatric and adult populations. Despite these refinements, the device still requires a bony recess of 3.5-4.5 mm depth (49).

The Osia employs a piezoelectric transducer, which produces mechanical vibrations when exposed to an electrical current. In contrast to the BONEBRIDGE, it is designed to sit atop the bone (not recessed in a well) and is anchored with an osseointegrated screw, thereby

minimising the extent of bone drilling. This design simplifies implantation and makes the device particularly suitable for children with thinner skulls. Despite its growing adoption, evidence in younger cohorts remains limited. The device was initially approved for patients 12 years and older in the United States (2019) and Australia (2022), before regulatory approval was extended in 2024 to include children aged 5-12 years, broadening its applicability and accessibility in the paediatric population.

The Sentio™ (Oticon Medical™, Askim, Sweden) is the most recent atBCI to enter the market in July 2024 (US) and May 2025 (Australia) for patients 12 years and over. Marketed by Oticon as the smallest atBCI currently available, its compact design offers potential advantages, though, like the BONEBRIDGE, it requires preparation of a bony recess. Given its recent release, evidence on its safety, audiological performance, and long-term outcome data has not yet been published.

Gaps in Knowledge

Although adult studies demonstrate a favourable safety profile and audiological outcomes with the Osia, evidence in younger children is sparse (44). Paediatric patients present unique anatomical and physiological challenges for implantable devices. Compared to adults, their thinner skull bones, softer tissue structures and ongoing craniofacial growth require meticulous surgical planning and long-term monitoring. In children with microtia-atresia, these challenges are further magnified. The associated anatomical abnormalities, such as underdeveloped mastoids, asymmetric craniofacial structures, and the need to plan for concurrent or future auricular reconstruction, can complicate implant positioning and secure anchoring of the transducer. There is therefore a need to review the current evidence regarding the safety and efficacy of paediatric Osia implantation.

This thesis aims to address these gaps by presenting the first systematic review of paediatric Osia outcomes and the first Australian case series of Osia implantation in children with microtia-atresia, providing valuable early insights into surgical safety, audiological benefit, and functional outcomes that can inform future larger-scale studies.

Chapter 2: Early Outcomes of Cochlear Osia Implantation in Children - A Systematic Review

Overview

This chapter presents a systematic review of published literature on paediatric outcomes of Osia implantation. Since its introduction in 2019, the Cochlear Osia has seen broad uptake in adults and, more recently, increasing use in children. Although this thesis is primarily concerned with hearing habilitation in children with microtia-atresia, very few studies to date have focused exclusively on this population, and even the broader evidence on paediatric Osia implantation remains limited. Nonetheless, children with aural atresia represent the majority of paediatric recipients – comprising over half of all reported cases in this review – making the available literature directly relevant to this group.

To date, no prior systematic review has specifically examined outcomes of Osia implantation in children, despite the unique anatomical, physiological, and developmental considerations that distinguish this population from adults. A review of paediatric outcomes is therefore warranted to address the gap in understanding of Osia performance and safety in younger patients.

The aim of this review is to summarise the current evidence on paediatric Osia implantation, with a focus on surgical technique, complications, audiological outcomes, patient-reported outcome measures and device compliance. This systematic review has been prepared as part of this thesis and is presented in manuscript form (in preparation for submission).

Title: Early Outcomes of Cochlear Osia Implantation in Children – A Systematic Review

Running title: Systematic review of paediatric Osia implantation

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Abstract

Background: The Cochlear™ Osia® is a newer active transcutaneous bone conduction implant increasingly used for children with conductive and mixed hearing loss and single-sided deafness. Although early studies in adults suggest promising outcomes, evidence in the paediatric population remains limited. This systematic review aims to summarise the current literature regarding Osia implantation in children.

Methods: A systematic search of MEDLINE, EMBASE and Scopus was conducted for studies published between 2019 and 2025 reporting outcomes of Osia implantation in patients aged ≤18 years. The review was performed in accordance with the PRISMA guidelines. Risk of bias was assessed using the Joanna Briggs Institute critical appraisal tools. Extracted outcomes included surgical complications, audiological outcomes, patient-reported outcomes (PROMs) and device wear time. Results were synthesised descriptively to accommodate variability in outcome measures and reporting.

Results: Eleven studies comprising 372 patients and 418 implants were included. Mean/median age at implantation was between 7.0 and 14.5 years. Most patients had conductive or mixed hearing loss (86.5%), most commonly due to aural atresia. Complications were uncommon. Minor soft-tissue complications were the most commonly described complication, although their reported frequency varied widely due to inconsistent definitions across studies. Major complications occurred in 2.2% of cases with an explantation rate of 1.3%. Weighted mean functional gain was 34.2 dB. Speech perception in quiet and noise improved significantly compared to unaided condition. PROMs showed statistically significant improvements post-Osia implantation. Overall device satisfaction, mean wear time and compliance were high.

Conclusion: Paediatric Osia implantation is associated with a favourable safety profile and clear audiological benefit, comparable to that seen in the adult literature. However, the

evidence base is limited to mainly small, heterogeneous observational studies with variable follow-up, highlighting the need for multicentre prospective research with standardised outcome measures and long-term follow-up.

Keywords: bone conduction, cochlear implants, hearing loss, systematic review, treatment outcomes

Introduction

Bone conduction implants (BCIs) are a cornerstone of hearing habilitation for children with microtia-atresia and are also widely used in managing conductive or mixed hearing loss (CMHL) from other causes, as well as single-sided deafness (SSD), in both paediatric and adult populations. BCIs function by converting sound into mechanical vibrations that are transmitted through the skull to stimulate the cochlea directly (50).

Surgically implanted BCIs are broadly classified as passive or active systems (Figure 1). *Passive* devices use an externally worn receiver-transducer that captures sound and converts it into vibrations, which are transmitted to an osseointegrated implant in the skull either percutaneously (via an abutment that passes through the skin - Figure 1D) or transcutaneously (via a magnet – Figure 1C). Percutaneous systems provide direct sound transmission to the bone but carry higher rates of wound complications and poorer osseointegration (51-54). Passive transcutaneous systems preserve intact skin and thereby reduce soft tissue complications; however sound transmission is attenuated by the interposed soft tissue between the actuator and magnet (1, 55). Furthermore, the retention magnet can put chronic pressure on the overlying skin, causing discomfort, skin ulceration or necrosis in severe cases. These issues can interrupt consistent device use during critical periods of learning and development in children (51).

Active transcutaneous BCIs (atBCIs) represent a newer generation of devices designed to overcome these limitations by maintaining an intact skin barrier while placing the transducer subcutaneously, thereby eliminating soft tissue attenuation and minimising skin-related complications (Figures 1A and B) (44, 55, 56). Owing to these advantages, atBCIs are increasingly supplanting passive devices in contemporary clinical practice.

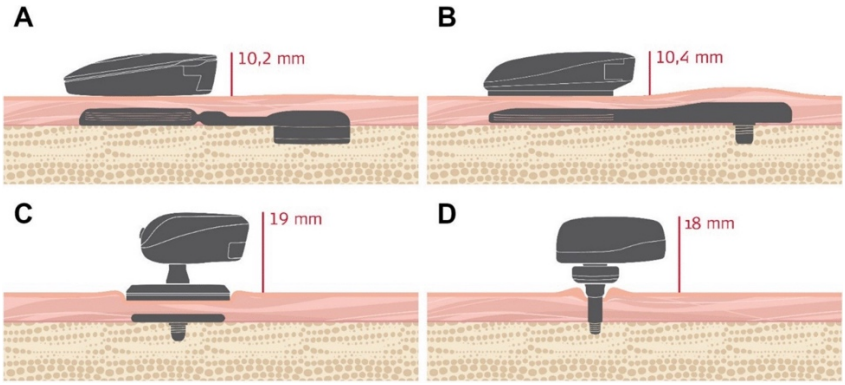

FIGURE 1. Active and passive bone conduction implants. A) Active transcutaneous implant with a floating mass transducer. B) Active transcutaneous implant with a piezoelectric transducer. C) Passive transcutaneous implant. D) Passive percutaneous implant; *from Caversaccio et al 2025* (42).

Three atBCIs are currently commercially available – the Osia® (Cochlear™ BAS, Gothenburg, Sweden), BONEBRIDGE® (MED-EL®, Innsbruck, Austria) and Sentio™ (Oticon Medical™, Askim, Sweden). The Osia has a unique technological profile, incorporating a ceramic piezoelectric transducer within a relatively thin 4.9 mm housing, and offering a fitting range of up to 55 dB HL. Because the device rests on the bone surface, it requires minimal (if any) bone removal to achieve a flat interface beneath the transducer. By contrast, both the BONEBRIDGE and Sentio utilise electromagnetic transducers that necessitate drilling a bony well - a step that can be challenging in young children with thin skulls (Figure 2). These devices also provide a narrower fitting range of up to 45 dB HL (47, 57).

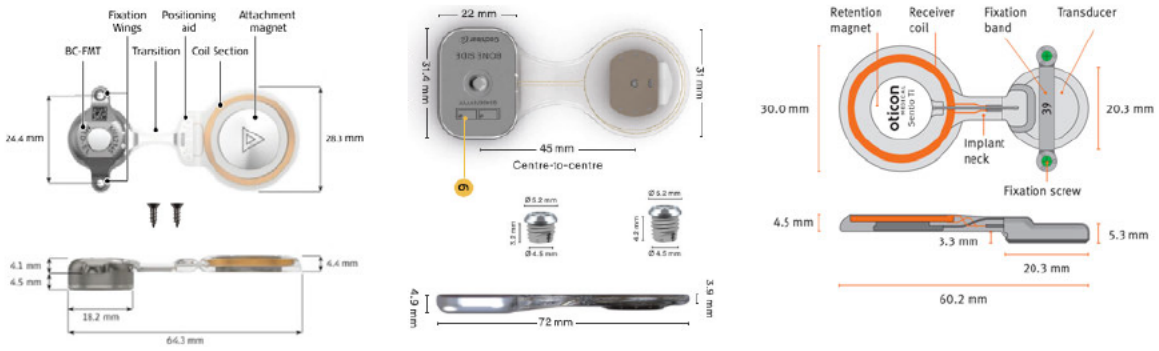

FIGURE 2. Active Transcutaneous Bone Conduction Implants. A) MED-EL BONEBRIDGE (BCI 602); *from MED-EL 2019* (58). B) Cochlear Osia (OSI300); *from Cochlear Ltd 2023* (59). C) Oticon Sentio (Ti implant); *from Oticon Medical 2025 (60)*.

The Osia was approved by the United States (US) Food and Drug Administration (FDA) and Australian Therapeutic Goods Administration (TGA) for patients 12 years and over in 2019 and 2022, respectively, and has shown promising safety and audiological outcomes in adult studies (44, 61). In 2024, the FDA and TGA expanded the indication to children five years and over. Importantly, the anatomy and indications for implantation in children differ substantially from adults. Paediatric candidates more frequently present with microtia and aural atresia, craniofacial syndromes, or other ENT-related comorbidities, and generally have thinner skulls, altered tolerability of external devices, and variable compliance (48). As a result, adult outcomes cannot be directly extrapolated to the paediatric population.

Despite the clinical importance of understanding paediatric outcomes with this new device, no review articles to date have synthesised the available evidence for the Osia in children. Paediatric data have begun to emerge over the past five years, with several case series and cohort studies reporting early surgical and audiological outcomes. This systematic review therefore aims to synthesise current evidence on the surgical safety and audiological outcomes of the Osia in patients ≤ 18 years of age. Secondary outcomes include patient or parent reported outcome measures (PROMs) and device compliance. We hypothesise that

Osia implantation in children has a low complication rate, significant audiological benefit and improved PROMs.

Methodology

Search Criteria

This systematic review was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) 2020 guidelines (62). A prospective protocol was developed prior to this review in accordance with the PRISMA-P statement (63). A comprehensive literature search was performed in MEDLINE, EMBASE, and Scopus for studies published between January 2019 (year of Cochlear Osia introduction) and 1 August 2025. The search combined MeSH-indexed search terms related to "Osia" and "Bone-anchored hearing implant" (Appendix 1). Reference lists of included studies and relevant literature reviews were also manually searched to identify additional eligible articles and confirm the search strategy.

Selection Criteria

All references were uploaded to systematic review data management software Covidence and screened for inclusion in this study. Duplicates were automatically removed. One reviewer (NN) screened all titles and abstracts for full text review, followed by two reviewers (NN and MS) performing independent full text review of selected articles against predefined inclusion and exclusion criteria. Discrepancies were resolved by discussion with a third reviewer.

The following PICOS criteria were used to determine study inclusion:

- Population – Children (≤18 years) with conductive, mixed or single-sided hearing loss
- Intervention: Osia (1, 2, or 3) implantation
- Comparison: None

- Primary outcomes: Intra- and post-operative complications and audiological outcomes
- Secondary outcomes: PROMs and device compliance
- Study design: randomised controlled trials (RCTs), cohort studies, case-control studies and case series with ≥5 patients.

Letters, editorials, commentary, case reports, case series with less than five patients, systematic reviews with no original data, animal, in-vitro, laboratory studies and non-English studies were excluded. Studies including adult patients were eligible only if ≥5 children were included in the study and paediatric outcomes were reported separately. Studies with a potential risk of cohort overlap (i.e. originating the same institution with partially overlapping or unclear recruitment periods) were not excluded provided each contributed unique outcome measures not reported elsewhere.

Data Extraction

Articles selected for inclusion underwent data extraction by two independent reviewers (NN, with either MS or NP), with discrepancies resolved by consensus or by a third reviewer. Extracted study characteristics included author, year of publication, funding, and country. Study design and level of evidence were classified according to established definitions from the Oxford Centre for Evidence Based Medicine (OCEBM) (64). Case series were defined as descriptive reports of patients undergoing the same intervention without a comparator, while cohort studies were defined as those systematically evaluating outcomes in a defined group of patients over time, either prospectively or retrospectively. Risk of bias (ROB) was assessed using the Joanna Briggs Institute (JBI) Critical Appraisal Checklists, applying the tool most appropriate to the study design (65).

Patient variables extracted included age, sex, ENT-related syndromes or comorbidities, type and aetiology of hearing loss, and prior hearing devices. Surgical data collected included implant laterality, operating time, incision type and location, soft tissue thickness, BI300 screw size used, and need for soft tissue reduction or bone polishing. Intraoperative complications and post-operative adverse events were also recorded. Audiometric outcomes were extracted as reported and included aided and unaided pure tone thresholds or pure tone averages (PTA), functional gain, speech testing in quiet and noise, sound localisation, and mean device wear time. Where available, PROMs were also collected.

Data Analysis

Continuous data (e.g. age, operating time, pure tone thresholds) were extracted and presented as reported in the original studies (means, medians, ranges, standard deviations). Where appropriate, sample-size-weighted means were calculated for descriptive synthesis across studies. However, true meta-analysis was not feasible owing to substantial heterogeneity in study design and outcome reporting, and results are therefore presented descriptively.

Results

Search Results and Study Characteristics

Of the 324 records identified, 194 unique studies were screened, 43 studies underwent full-text review, and 11 studies met inclusion criteria (Figure 3). Study characteristics are presented in Table 1. Publications spanned 2022 to 2025 and originated from four countries – the United King, the United States, Canada and Saudi Arabia. Five were classified as cohort studies (OCEBM Level III) and six as case series (Level IV). Six were conducted prospectively and five retrospectively (64). Overall, risk of bias across included studies was rated as low to moderate. Most studies clearly defined inclusion criteria, demographics, and used consistent outcome measures; however methodological quality was limited by small

sample sizes, heterogeneous follow-up, variable reporting of comorbidities and other confounders, and retrospective design in a subset of studies (Appendix 2).

Across the 11 included studies, 372 patients underwent a total of 418 Osia implants. Median study size was 25 patients (range 7-124 patients). Mean or median follow-up was reported for six studies. Three had follow-up periods of less than 12 months (range 5.3-12 months) (66-68), while the other three reported longer-term follow-up ranging from 24 to 35 months (69-71) (Table 1).

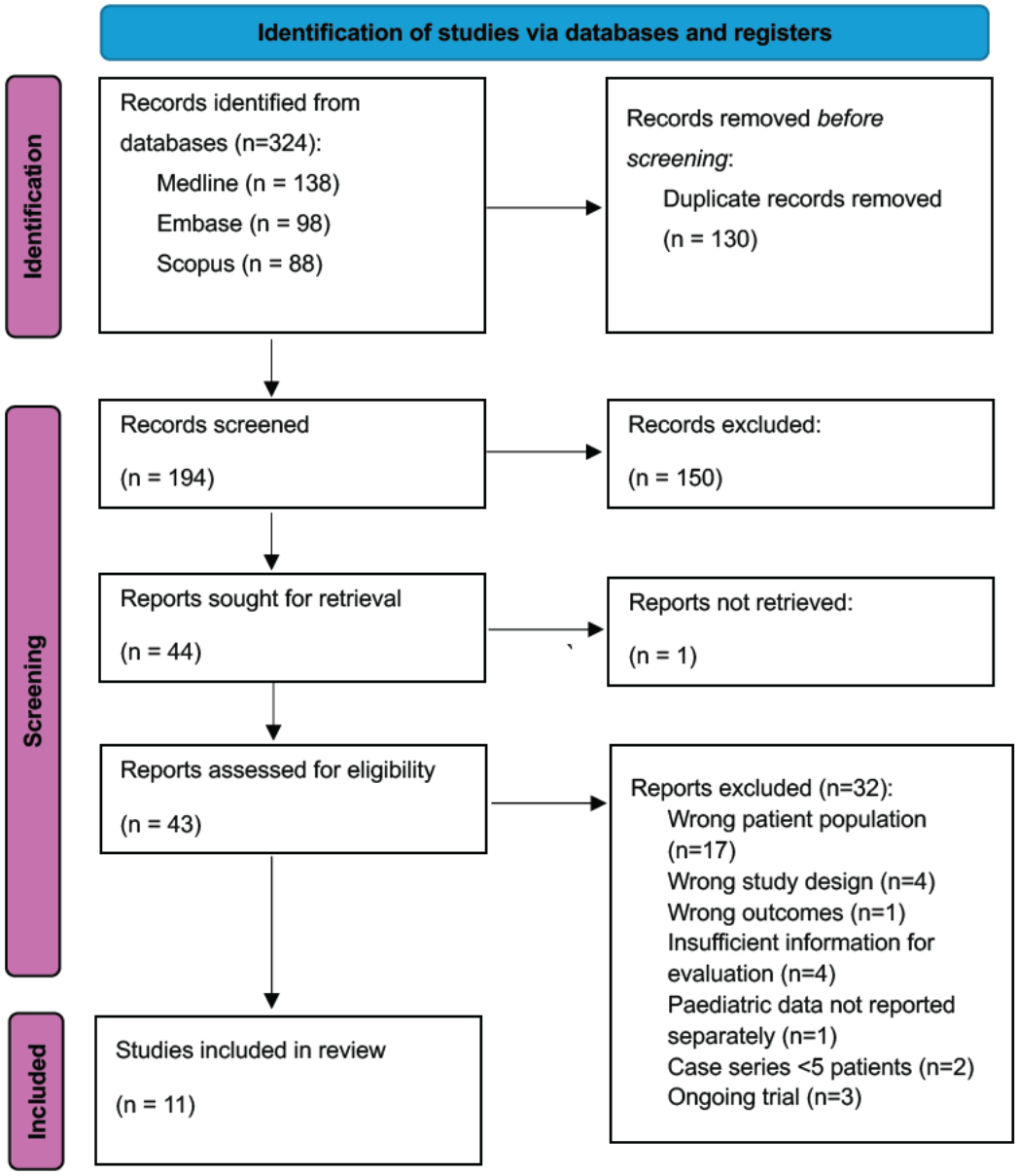

FIGURE 3. PRISMA Flow Diagram

TABLE 1. Study Characteristics with Associated Outcomes

	Study Characteristics				Baseline Characteristics				Outcomes			
Author	Country	Design	Timing	LOE	Patients (n)	Mean Age (Range)	Female (%)	Hearing loss type	Complications reported	Audiological Outcomes	PROM	Mean follow-up (Mo)
Mozaffari 2025 (72)	UK	Cohort (single-arm)	R	III	49	9.6 (5.2-18.8)*	46.9	All	Yes	Mean pure tone thresholds, SIN	NR	NR
You 2022 (66)	USA	Case series	R	IV	16	12.9 (10.2-17.6)	68.8	CMHL	Yes	Mean PTA, FG, SRT (quiet), WRS (quiet)	NR	7.1 ± 4.0
Florentine 2022 (67)	USA	Case series	R	IV	14	11.5 (7.4-15.6)	28.6	All	Yes	Mean PTA	NR	5.3 ± 3.7
Alnoury 2023 (73)	Saudi Arabia	Case series	P	IV	7	13 (6-17)	NR	NR	Yes	NR	NR	NR
Stevens 2025 (68)	USA	Cohort (single-arm)	P	III	50	7.4 (5-11)	54.0	All	Yes	Mean PTA, WRS (quiet), SIN	SSQ	12**
Zawawi 2025 (70)	Saudi Arabia	Cohort (single-arm)	P	III	23	7 (3-17)*	52.2	CMHL	Yes	Median PTA, WRS (quiet)	NR	24.23 (IQR 15.47)*
Zawawi 2025 (69)	Saudi Arabia	Cohort	P	III	25	7 (3.1-17.5)*	52.0	All	Yes	Median PTA, WRS (quiet)	HEAR-QL	34.83 (IQR 15.78)*
Cushing 2022 (74)	Canada	Case series	P	IV	42	10.9 (4.9-18)	50.0	All	Yes	NR	NR	NR
Cushing 2024 (75)	Canada	Cohort	R	III	124	11 (4.9-18)	48.4	All	Yes	NR	NR	NR
Gordon 2022 (76)	Canada	Case series	P	IV	14	14.5 (13.3-15.8)*	57.1	All	Yes	Mean PTA, WRS (quiet)	SSQ	NR
Leonard 2025 (71)	USA	Case series	R	IV	8	9 (5-15)	NR	CMHL	Yes	NR	NR	26

* = median rather than mean reported; ** = fixed study endpoint; CMHL = Conductive or Mixed Hearing Loss; FG = Functional Gain; HEAR-QL = Hearing Environments and Reflection on Quality of Life; LOE = Oxford Centre of Evidence Based Medicine Level of Evidence; Mo = months; NR = Not Reported; P = Prospective; PROM = Patient-Reported Outcomes Measures; PTA = Pure Tone Average; R = Retrospective; SIN = Speech-in-noise; SRT = Speech Reception Threshold; SSQ = Speech, Spatial and Qualities of Hearing Scale; WRS = Word Recognition Score

Patient Characteristics

Mean or median age was reported in all studies and ranged from 7.0 to 14.5 years, with an overall age span of 3.0-18.8 years. Sex distribution was approximately equal (50.4% female). Five studies (67, 70, 72, 74, 75) detailed the presence of ENT-related syndromes or comorbidities, which affected 32.5% of patients. The most common were craniofacial syndromes such as Goldenhar syndrome, Treacher Collins syndrome, and hemifacial microsomia. Other reported conditions included Pierre Robin sequence, CHARGE syndrome, Trisomy 21 and 22, chromosome 18q deletion, craniosynostosis, Apert syndrome, Branchio-Oto-Renal syndrome, microcephaly, Negar syndrome and oculoectodermal syndrome (Table 2).

TABLE 2. Patient Characteristics

Sex	*n = 357*	*%*
Female	180	50.4
Male	177	49.6
ENT-related syndromes or comorbidities	*n = 252*	
Yes	82	32.5
No	170	67.5
Type of hearing loss	*n = 365*	
CMHL	312	85.5
SSD	53	14.5
Aetiology of hearing loss	*n = 339*	
Aural atresia	234	69.0
Canal stenosis	40	11.8
Cochlear nerve hypoplasia	23	6.8
Other/unknown	14	4.1
Unspecified SSD	8	2.4
Middle ear dysplasia	7	2.0
Chronic middle ear disease	7	2.0
Cochlear anomalies	6	1.8
Previous hearing device use	*n = 245*	
Yes	228	93.1
No	17	6.9
Implant laterality	*n = 372*	
Unilateral	326	87.6
Bilateral	46	12.4

CMHL = conductive or mixed hearing loss; SSD = single-sided deafness

Of studies reporting hearing loss type, the majority of patients had conductive or mixed hearing loss (CMHL) (85.5%) compared with single-sided deafness (SSD) (14.5%). Four studies included only patients with CMHL (66, 70, 71, 77), while the remainder included patients with SSD. Hearing loss aetiology was described in nine studies on a per-patient basis, with the most common cause being aural atresia (69.0%), followed by canal stenosis (11.8%) and cochlear nerve hypoplasia (6.8%). Across five studies, most children (93.1%) had prior experience with hearing devices (Table 2) (66, 72, 74-76).

Surgical Technique

Unilateral implantation was most common (87.6%), with three studies including only unilateral cases (68, 71, 73) and the remainder including both unilateral and bilateral procedures. Mean or median operating time, reported in seven studies, ranged from 34 to 87.2 minutes (66, 68-71, 74, 76).

Incision type and position varied among the ten studies that described surgical technique, but was often tailored to whether microtia reconstruction or other concurrent surgery was planned, with several groups highlighting the importance of preserving vascular supply for future reconstruction (67, 72). Anteriorly based (relative to device) post-auricular incisions were generally preferred in children with a typical pinna and posteriorly based scalp incisions preferred in children with microtia or atresia (66, 67, 70, 72, 74-76). Four studies emphasised maintaining a soft tissue clearance of at least 10-15 mm between the edge of the actuator and incision to avoid skin tension, in keeping with manufacturer recommendations (66, 70, 74, 76).

Non-standard implantation techniques were reported in three studies. Alnoury et al (73) described a minimally invasive endoscopic-assisted technique using a transverse skin incision over the actuator, with biplane dissection at two tissue levels to prevent device exposure in case of dehiscence. Leonard et al (71) reported their approach for simultaneous Osia implantation and alloplastic ear reconstruction using a temporoparietal or occipital fascial flap, which involved a larger periauricular incision to allow flap harvest and inset, with the Osia positioned posteriorly. Zawawi et al compared outcomes of BI300 screw placement at 5.5cm (Group 1) versus 7cm (Group 2) from the external auditory canal in children with microtia, with group allocation determined by feasibility of placement based on prior reconstruction and likelihood of future reconstruction (70).

Soft tissue thickness overlying the receiver was reported in five studies, with mean or median soft tissue thickness ranging from 3.5 to 6.3 mm (67-70, 76). Three studies reported rates of bone bed polishing with a drill to achieve adequate clearance around the actuator. Florentine et al (67) reported bone polishing was required in 1/43 implants (2%), You et al (66) in 4/20 implants (20%), and Stevens et al (68) in 18/50 implants (36%). BI300 screw size was reported in four studies, with 3 mm screws used more frequently (n = 107/177 ears, 60.5%) than 4 mm screws (n = 70/177 ears, 39.5%) (67, 68, 72, 74).

Intra-operative Events

Among the four studies (n = 150 patients) that reported on intra-operative complications, there were four bleeding incidents reported (one emissary vein, three sigmoid sinus), which were controlled intra-operatively (68, 69, 72, 73). There were 26 reports of dural exposure across two studies - 1/7 in one study (73) and 25/50 in the other (68) - none of which resulted in dural injury or CSF leak. Cushing et al, who utilised a 4 mm BI300 screw in all cases,

noted that dural exposure was common (frequency not reported) but did not result in dural injury (74). There were two reports of the drilling site being changed due to inadequate skull thickness (69).

Adverse events

Reported adverse event rates ranged from 0% to 34% across studies. None of the included studies used a standardised surgical complication grading framework such as the Clavien–Dindo Classification System (78), and the absence of uniform severity grading limits comparability across studies and likely contributes to the wide variability in reported adverse event rates. Due to heterogeneity in adverse event definitions and reporting practices, a pooled summary statistic for all-severity complications could not be calculated.

Minor or expected post-operative issues – corresponding broadly to Clavien-Dindo Grade I or II events - were inconsistently reported. These included transient post-operative pain, erythema or skin irritation, swelling, small seromas, headache, and minor infections (66-70, 72-75). Such events were typically self-limiting or resolved with conservative management, although inconsistencies in how they were documented meant that their true incidence could not be reliably compared across studies. Some studies documented them in detail, while others likely omitted them as anticipated or clinically insignificant.

Major complications - defined as those requiring return to surgery, device explantation or device non-use - largely represented Clavien-Dindo Grade III events. These occurred in 2.2% of cases (Table 3) and included five cases of device explantation due to infection or device exposure (66, 75), one case of magnet retention issues requiring repeated skin-thinning procedures in an adolescent with obesity and BOR syndrome (75), one post-operative

haematoma requiring surgical exploration and washout (68), and one case of pressure-related pain resulting in device non-use (66). At least four of the five explants occurred in children with complex comorbidities or surgical history, including two children, aged five and nine, with trisomy 21 (one with a previously extruded/infected percutaneous implant), a 15-year-old with Smith-Magenis syndrome with a prior percutaneous abutment removed for recurrent infection, and a 16-year-old with prior rib cartilage microtia reconstruction and a history of an extruded magnet from a previous passive device (75). In their series on concurrent Osia implantation and alloplastic auricular reconstruction, Leonard et al reported one return to surgery for revision of the helical rim overlying the auricular reconstruction, however no Osia-related complications were observed (71).

No Clavien-Dindo Grade IV (life-threatening) or Grade V (death) complications were observed.

TABLE 3. Adverse Events

Intra-operative complications	*n = 150*	%
Sigmoid sinus bleeding	3	2.0
Emissary vein bleeding	1	0.7
Major complications	*n = 372*	
Device explantation	5	1.3
Magnet retention issues requiring repeated skin thinning	1	0.3
Post-operative haematoma requiring washout	1	0.3
Device non-use	1	0.3

Audiological outcomes

Audiological outcomes, reported in seven studies, consistently showed significant improvement with Osia (66-70, 72, 76). Change in mean or median PTA was the most commonly reported metric, though method of audiometric data collection, PTA calculation, and outcome reporting was highly variable, limiting pooled analysis (Appendix 3). Three studies (66, 68, 76) reported paired unaided and Osia-aided mean PTAs using 0.5-4 kHz

thresholds (n = 79 patients), with a weighted mean functional gain of 34.2 dB (range 27.9-43.1 dB). A further two studies (69, 70) reported median PTAs using 0.25-8 kHz thresholds (n = 47 patients), with improvements from 60-67.5 dB HL unaided to 20-21.67 dB HL with Osia.

Comparisons with other hearing devices were limited to three studies. Collectively, these studies suggested superior high-frequency performance and overall aided thresholds with Osia. Florentine et al found similar mean aided PTAs between various non-Osia devices (25.1 ± 7.7 dB HL) and Osia (26.4 ± 4.9 dB HL), but Osia preserved high-frequency gain (8 kHz threshold 23.8 ± 7.5 dB HL vs 42.5 ± 10.6 dB HL) (67). Stevens et al reported significantly better aided thresholds with Osia compared to the Baha 5 Power Softband (23.1 ± 5.4 vs 27.6 ± 6.9 dB HL; p<0.0001) (68). Similarly, Mozaffari et al noted threshold improvements at multiple frequencies, reaching statistical significance at 4 kHz ($p < 0.0001$) (72).

Speech Testing

Speech audiometry was assessed in five studies using a variety of tests in both quiet and noise, all demonstrating substantial improvement with Osia. In quiet, You et al reported reduction in SRT from 63.2 ± 12.6 dB HL unaided to 22.1 ± 4.9 dB HL with Osia (66). Stevens et al observed median CNC word scores rising from 14% unaided to 86% Osia-aided, with Osia also outperforming Baha 5 Power Softband (86% vs 72%) (68). Zawawi et al similarly demonstrated marked gains in speech discrimination from 35% to 90% in CHL and from 3% to 86% in SSD (both $p \leq 0.001$) (69). In their study comparing implant placement positions, speech discrimination was similarly high (>90%), with no significant differences across placement groups (70).

Speech-in-noise (SIN) outcomes, reported by two studies, also supported Osia benefit. Mozaffari et al found significant Osia-aided improvements at 0, +5, and +15 dB signal to noise ratio (SNR) compared with unaided conditions, with the strongest effect at 0 dB SNR ($p = 0.042$), representing the most challenging listening conditions where speech and noise are at equal intensity (72). Using the Bamford-Kowal-Bench SIN test, Stevens et al found a mean Osia-aided improvement in SNR for 50% correct speech recognition (SNR-50) of -10.1 ± 7.8 dB compared with unaided listening (mean of differences, $p<0.0001$) (68).

Sound localisation was assessed in two studies. In one, median accuracy for determining sound laterality improved from 34-38% unaided to 76-82% Osia-aided at six months post-operatively ($p = 0.004$) (70). In the other, mean accuracy in children with CHL increased from 36% unaided to 76% Osia-aided ($p = 0.004$), while in SSD children accuracy rose from 15% to 32% ($p = 0.04$) (69).

Patient-Centred Outcomes: PROMs, Qualitative Feedback, and Device Use

Three studies collected PROMs using validated questionnaires. Two utilised the Speech, Spatial, and Qualities of Hearing Scale (SSQ), which evaluates listening ability in a range of everyday contexts, and one employed the Hearing Environments and Reflection on Quality of Life (HEAR-QL) survey, a measure of self-perceived hearing-related quality of life in children (34, 79). Across the two SSQ studies, scores improved significantly at 6 months post-operatively, with a weighted mean increase of 1.73 points (range 1.63-2.1 points; $p < 0.0001$ for both studies) (68, 76). In the HEAR-QL study, scores improved from 68% pre-operatively to 87% with Osia at both 6 months and 2 years follow-up ($p = 0.001$) (69).

Three studies summarised patient and caregiver perspectives. Overall satisfaction was high, with families reporting improved compliance, communication, learning, and behaviour (72, 73). In one study, parents of children with neurosensory disorders reported particular benefit, with Osia better tolerated than softband devices (72). Patients described superior sound quality and comfort compared to prior passive devices, with all children with previous device experience preferring Osia in one study (66).

Device wear time was reported in two studies, with mean or median daily duration of use ranging from 9.8 to 10.3 hours at 6 months post-operatively (68, 70). Some practical challenges were noted relating to magnet strength, with some patients requiring stronger magnets than initially recommended for secure retention (66). One study reported a median retention score of 8/10 at 6 months (95% CI 8-9), with a slightly lower retention rating in sports (median 7/10; 95% CI 6-8).

Discussion

This is the first review to specifically evaluate paediatric outcomes of the Osia, synthesising the initial published experience of implantation in children. The focus of this review on the Osia reflects both its recent TGA approval for children five years and over and its increasing adoption in paediatric clinical practice, particularly among children with microtia-atresia and other complex comorbidities. Despite this growing clinical uptake, outcome data in children remain limited, underscoring the value of synthesising early results.

In this review, the majority of patients (86.5%) had CMHL, with aural atresia being the most common indication for surgery (58.5%). Other systematic reviews of predominantly adult Osia cohorts have reported similar rates of CMHL (~80%), but with different surgical

indications: aural atresia accounted for a smaller proportion of cases (27.1%), while chronic ear disease was more frequently reported (22.3% vs 2% in this review) (44, 61). Among paediatric studies reporting the presence of ENT-related syndromes or comorbidities, almost one-third of children were affected. These differences highlight that the paediatric Osia population may have distinct baseline characteristics compared with adults, particularly with respect to underlying anatomy and associated conditions - an important consideration when interpreting outcomes and their generalisability.

Surgical technique varied across studies, reflecting the evolving nature of paediatric Osia implantation and the lack of a standardised approach for children. Variability in incision type, actuator position, soft-tissue thickness management, and bone bed preparation may plausibly influence both audiological performance and soft-tissue related complications. Although few studies directly examined technique-outcome relationships, this heterogeneity may contribute to variability in reported outcomes and should be considered when interpreting the literature. Future studies comparing specific techniques or providing more granular reporting may help clarify their impact.

Outcome reporting across studies was highly variable, particularly in the definition and documentation of adverse events. Intra-operative issues such as sigmoid sinus and dural exposure were inconsistently reported, likely stemming from the fact that these are variably regarded as complications. In two studies that did report on it, high rates of dural exposure were acknowledged, though without associated sequelae, consistent with the literature (68, 74). Major complications were more consistently captured, with a low overall rate of 2.2% and explantations in 1.3% of cases. These findings are broadly consistent with systematic reviews of predominantly adult Osia cohorts, which have reported major complication rates

of around 1% (61). Importantly, at least four of the five explants in this review occurred in children with complex comorbidities or a history of previous device infections or extrusions. This suggests that higher complication rates in children may be concentrated among those with syndromic diagnoses or previous implant complications, underscoring the importance of careful surgical selection, tailored pre-operative counselling, vigilant wound surveillance by caregivers and close post-operative follow-up in this subgroup.

Audiological outcomes were reported with considerable variability across studies (Appendix 3), likely reflecting heterogeneous audiometric testing protocols between centres, differences in age and developmental level affecting children's ability to participate reliably in formal testing, and the retrospective nature of many studies, which limited standardisation of reported outcomes. Despite this, results were consistently favourable. Statistically significant improvements in mean PTA were observed, with a weighted mean functional gain of 34.2 dB across three studies. This aligns closely with published literature, with Key et al reporting mean functional gain of 35 dB (95% CI 29.12-40.97) in their systematic review (44). The Osia also consistently outperformed passive bone conduction devices, with three studies highlighting preservation of high-frequency gain. Speech audiometry likewise showed consistent post-operative benefit in both quiet and noise, with speech discrimination scores improving from poor unaided performance to over 80% in most cases. Again, outcomes with Osia exceeded passive systems, likely reflecting improved access to high-frequency consonants that underpin speech clarity.

This finding is echoed in several atBCI studies, largely attributed to their ability to bypass soft-tissue attenuation (55, 56, 67, 68, 72, 80-82). Some comparative reports between the Osia and electromagnetic atBCIs have suggested superior gain with the Osia, potentially

reflecting the capacity of its piezoelectric actuator to generate higher sound pressure levels at high frequencies than electromagnetic systems. However, many of these comparisons are limited by small sample sizes and heterogeneity in patient groups, which restricts the strength of the conclusions (83-85). This remains an important area for future research.

Validated PROMs were collected in only three studies using the SSQ or HEAR-QL inventories, all of which showed statistically significant improvement following Osia implantation (68, 69, 76). The inclusion of PROMs is particularly important in paediatric cohorts, where comprehensive audiometric testing can be limited by age, attention, cognitive ability and behavioural factors (72, 86). Beyond serving as a surrogate where formal testing is challenging, validated questionnaires provide complementary insights into listening abilities and communication across real-world settings. This is particularly relevant in children with unilateral hearing loss, where the benefits of intervention may not be fully reflected in conventional thresholds but are captured in functional or quality-of-life domains such as localisation, classroom listening and fatigue (86). Including PROMs in future studies will therefore enhance the assessment of broader functional and quality-of-life benefits of hearing devices in children.

This systematic review is subject to several important limitations that should be considered when interpreting its findings. The overall evidence base for paediatric Osia implantation remains limited to observational studies, primarily single-centre case series and cohort studies, corresponding to Oxford Levels of Evidence III-IV (64). No Level I-II studies (systematic reviews or randomised controlled trials) were identified. The predominance of early feasibility and pilot studies raises the risk of publication bias, as positive results are more likely to be published, while negative or null findings may be underreported. Cohort

sizes were modest, ranging from 8 to 124 patients, which is not unexpected given the very recent approval of Osia implantation in children under 12 years. Several included studies therefore reported outcomes from “off-label” use. Despite these constraints, quality appraisal identified a subset of well-designed prospective trials with clear methodology and robust outcome reporting (Appendix 2) (68-70, 87).

Marked heterogeneity was observed in outcome measures across studies as discussed above, which prevented quantitative pooling beyond descriptive synthesis. Most studies provided only short- to medium-term follow-up, restricting conclusions about device durability and sustained benefit. Several studies included wide age ranges from early childhood to late adolescence, introducing variability in skull thickness, soft-tissue characteristics, developmental stage, and engagement with rehabilitation. These factors influence surgical safety and device tolerability. Finally, some studies - (69, 70) and (74-76) - originated from the same institutions and may include overlapping cohorts, raising the potential for double-counting of outcomes. These studies were retained in the review for two reasons. First, each contributed unique outcome measures (e.g. audiological outcomes, PROMs, surgical variables) not reported elsewhere, thereby enriching the overall synthesis. Second, given the limited number of paediatric Osia studies available to date, exclusion would have further reduced the already small evidence base and risked omitting potentially valuable data. Use of narrative synthesis rather than meta-analysis mitigates the risk of bias, and the presence of overlapping cohorts is transparently acknowledged as a limitation.

Going forward, future research should prioritise high quality, multicentre prospective studies with larger sample sizes to strengthen the evidence base. Standardisation of outcome reporting is critical, particularly for audiological and speech measures, where consistent PTA

frequency ranges, speech-in-quiet, and speech-in-noise protocols, and validated PROMs should be employed to better evaluate hearing outcomes in real-world listening environments and enable comparison across studies. Long-term follow-up is also needed to assess device durability, complication rates, and sustained impact on hearing, quality of life and educational outcomes. Stratifying outcomes by surgical technique, co-morbidities, and age - particularly isolating younger children, where thin skulls and soft-tissue fragility are most relevant - would strengthen interpretation of complication rates and audiological benefit. Comparative studies between Osia, BONEBRIDGE and Sentio would help clarify relative benefits and risks, particularly in young children, those with craniofacial syndromes, and those undergoing simultaneous reconstruction where differences in device size, profile and implantation technique are of particular relevance. Finally, development of international registries or collaborative databases may help overcome small cohort sizes, reduce reporting bias, and provide more generalisable insights into outcomes in this population.

Conclusion

Early paediatric experience with the Osia system highlights substantial audiological and speech perception benefits for children with CMHL and SSD, with a low major complication rate. Many of the challenges associated with translating adult outcomes to paediatric populations – such as thinner skulls, concurrent reconstructive surgery, craniofacial syndromes, and differences in compliance and device tolerability – are highlighted by this evidence base. However, the current evidence is limited by small, heterogeneous observational studies, and findings should therefore be interpreted with caution.

Although long-term paediatric Osia data are limited, early patterns suggest that audiological gains and device tolerability are likely to remain stable, consistent with outcomes from earlier

bone-conduction systems. Major complications appear most common in the early postoperative period, with few late adverse events reported in analogous devices. However, true 5- and 10-year outcomes - particularly regarding implant longevity, soft-tissue health, evolving soft-tissue thickness with growth, and sustained functional benefits - cannot yet be determined. In the paediatric population, increasing soft-tissue thickness through adolescence and adulthood may influence long-term magnet retention or necessitate revision surgery, but these effects have not yet been observed. Larger, standardised and long-term studies are required to confirm these benefits, clarify the impact of age-related soft-tissue changes, and better inform clinical decision-making in paediatric hearing rehabilitation.

Chapter 3: Outcomes of a New Active Transcutaneous Bone Conduction Implant in Children with Microtia-Atresia – First Australian Experience

Overview

This chapter presents a retrospective case series of ten Osia implants performed in nine children aged 12 years and under with microtia-atresia. Most children in this study underwent concurrent single-stage alloplastic auricular reconstruction with a 3D-printed porous polyethylene framework, a novel approach that reflects the close integration of hearing habilitation and cosmetic reconstruction in this population. The chapter reports practical considerations for a combined approach in this population, as well as reporting on intra- and post-operative complications, audiological outcomes and functional listening performance using the Parents' Evaluation of Aural/Oral Performance of Children (PEACH) questionnaire. To the author's knowledge, this study represents the first Australian experience of paediatric Osia implantation, and only the second international report of simultaneous Osia implantation with single-stage alloplastic auricular reconstruction. This case series has been prepared as part of this thesis and is presented in manuscript form (in preparation for submission).

Title: Outcomes of a New Active Transcutaneous Bone Conduction Implant in Children with Congenital Aural Atresia – First Australian Experience

Running title: Paediatric Outcomes of a New Active Transcutaneous Bone Conduction Implant

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Abstract

Introduction: Congenital microtia-atresia causes moderate to severe conductive hearing loss. Active transcutaneous bone conduction implants (atBCIs), such as the Cochlear™ Osia®,

offer an alternative to traditional passive bone conduction devices, but evidence in children is limited. The Osia was only recently approved in Australia for children aged 5-12 years (2024), and no Australian paediatric outcome data have been reported.

Methods: A retrospective case series of children with microtia-atresia who underwent Osia implantation between 2022 and 2025 was conducted. Data included demographics, surgical technique, complications, audiological outcomes, Parents' Evaluation of Aural/Oral Performance of Children (PEACH) scores and qualitative feedback.

Results: Nine children with microtia-atresia (10 implants) were included (median age 8.8 years; range 4.8-12.8; 60% male). Eight implants were performed with concomitant alloplastic auricular reconstruction with a 3D-printed porous polyethylene framework. Two devices required explantation due to skin breakdown, with both complications occurring in patients with hemifacial microsomia undergoing concurrent auricular reconstruction. Audiological testing demonstrated markedly improved Osia-aided thresholds compared to unaided conditions. PEACH scores improved in quiet and in noise, with a median global improvement of +10%. Parents consistently reported improved compliance, reduced listening fatigue and better school and social participation compared to previous devices.

Conclusion: Osia implantation is feasible in younger children with microtia-atresia and provides consistent audiological and functional benefit. No bone-related complications were observed; however, soft tissue complications were more common in children with hemifacial microsomia undergoing concurrent microtia reconstruction, likely due to the presence of thin overlying skin in the setting of temporoparietal fascial flap harvest. These findings represent the first Australian paediatric Osia outcomes and underscore the need to balance the benefits of early auditory rehabilitation with careful device selection, particularly as more devices become approved for children. Multicentre studies with longer follow-up are warranted to further evaluate long-term safety and effectiveness.

Keywords: Congenital aural atresia, hearing loss, active transcutaneous bone conduction implants, case series

Introduction

Children with congenital microtia-atresia face two major challenges: the functional burden of conductive hearing loss and the psychosocial impact of a visible craniofacial difference. Microtia-atresia affects 0.83 to 17.4 per 10,000 births, is unilateral in 77-92% of cases and typically results in a moderate to severe conductive hearing loss due to absent or stenotic external and middle ear structures (1, 2). Between 20% and 60% of affected children present with associated anomalies or syndromes, such as hemifacial microsomia, Goldenhar syndrome, Treacher Collins syndrome and Trisomy 21 (3). In addition to impaired auditory access, up to 90% of affected children experience social stigmatisation, stemming from both the visible malformation and from the hearing disability itself, which contributes to listening fatigue, impaired socialisation, and reduced confidence. These challenges underscore the importance of early interventions that address both hearing habilitation and auricular reconstruction (88, 89).

Conventional management options include atresiaplasty, adhesive or soft-band retained bone conduction devices, and passive bone-anchored hearing implants. Eligibility for atresiaplasty depends on temporal bone anatomy and audiometric results, and only around 50% of patients meet the criteria (37). Furthermore, audiological outcomes following atresiaplasty are variable, and most children require ongoing use of hearing devices. Lifelong aural care is typically necessary, with a high risk of restenosis (up to 30% in the literature) and other complications, and revision surgery may be necessary (38, 39).

Passive bone conduction devices, while less invasive, have been associated with soft tissue complications (particularly percutaneous devices), frequent feedback, limited high-frequency transmission, and poor compliance in children (38, 66). These limitations have driven interest in active transcutaneous bone conduction implants (atBCIs), which maintain intact skin and avoid soft tissue attenuation by housing the transducer internally. Currently available atBCIs include the BONEBRIDGE® (MED-EL, Innsbruck, Austria) and the Osia® (Cochlear™ BAS, Gothenburg, Sweden). The Sentio™ (Oticon Medical AB™, Askim, Sweden) has also recently been introduced (2025) but is not currently approved for use in children under 12 years of age in Australia (Figure 1).

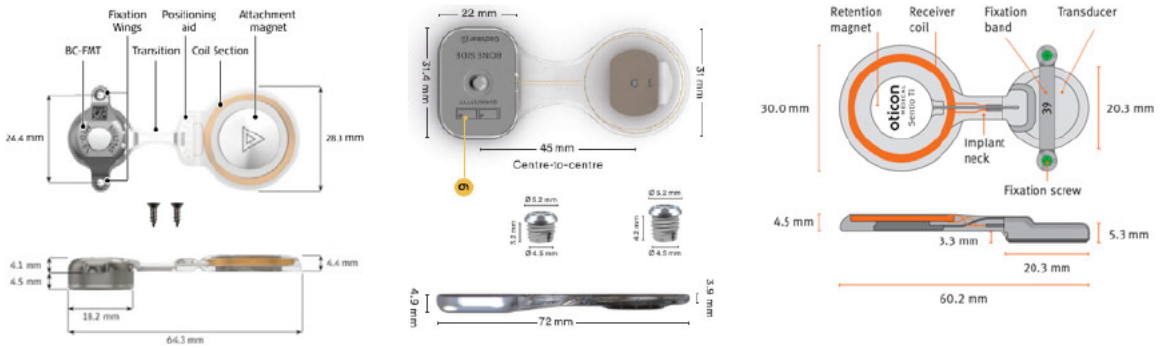

FIGURE 1. Active Transcutaneous Bone Conduction Implants. A) MED-EL BONEBRIDGE (BCI 602); *from MED-EL 2019* (58). B) Cochlear Osia (OSI300); *from Cochlear Ltd 2023* (59). C) Oticon Sentio (Ti implant); *from Oticon Medical 2025 (60).*

The Osia employs a piezoelectric actuator and requires minimal if any bone removal. It was approved by the Therapeutic Goods Administration (TGA) for use in patients 12 years and over in Australia in 2022 and extended to children aged 5-12 in 2024. Adult series have reported favourable safety, audiological, and patient-reported outcomes, but paediatric data remain limited to mainly small observational studies (44). Importantly, no Australian paediatric outcome data have yet been published.

Parallel advances in microtia reconstruction have also shifted management. Whereas traditional autologous rib graft reconstruction is typically deferred until seven to ten years of age to ensure adequate donor cartilage, newer prefabricated alloplastic implants made of porous polyethylene allow reconstruction in children as young as four (90-92). This shift has created momentum to consider hearing habilitation at a younger age, allowing reconstruction and implantation to be performed in a single procedure.

In such cases, a vascularised temporoparietal fascia flap pedicled on the superficial temporal artery provides robust coverage for the alloplastic auricular framework and simultaneously allows placement of a sizable atBCI without a separate skin incision. This approach provides the advantage of protecting the bulk and metallic components of the device beneath thick, vascularised tissue and away from the incision. However, a disadvantage is that the superior part of the device containing the magnet often sits under thin skin, as the temporoparietal flap is rotated inferiorly to cover the auricular framework. Regardless, such an approach not only avoids multiple staged surgeries but also provides earlier access to consistent auditory input during critical developmental periods (93). To date, only one case series describing single-stage Osia implantation and alloplastic auricular reconstruction has been described in the literature (71).

This study presents the first reported Australian experience with Osia implantation in children 12 years and under with microtia-atresia, the majority of whom underwent simultaneous alloplastic auricular reconstruction with a 3D-printed porous polyethylene implant. The aim of this study was to evaluate the surgical safety, audiological benefit, and parent-reported functional outcomes of Osia implantation in younger children with microtia-atresia, particularly in the context of combined microtia reconstruction.

Methodology

Study design and setting

A retrospective case series of consecutive paediatric microtia-atresia patients who underwent Cochlear™ Osia® implantation between 2022 and 2025 was conducted. All procedures were performed by a single otologist in collaboration with a single plastic surgeon for cases with combined auricular reconstruction. The study was approved by the Royal Prince Alfred Hospital Human Research Ethics Committee (reference X25-0154).

Participants

Children were eligible for inclusion if they:

1. Had unilateral or bilateral microtia-atresia
2. Were ≤12 years at the time of surgery, and
3. Received at least one Osia implant during the study period.

Exclusion criteria were no available post-operative outcome data or less than three months of follow-up.

Surgical technique

All patients underwent pre-operative high-resolution computed tomography of the petrous temporal bones (CT PTB) to determine bone and soft tissue thickness and guide implant placement.

In combined cases, the ENT and reconstructive plastic surgeons jointly marked the planned implant, incision and temporoparietal fascia (TPF) flap sites (Figure 2). A curved post-auricular incision was made to facilitate auricular framework placement. Subcutaneous dissection was carried out up to the superior temporal line to facilitate harvest of the

temporoparietal fascia flap (TPF), with preservation of the periosteum. The alloplastic auricular implant was then secured, and the TPF flap rotated over the framework to ensure adequate vascularised coverage.

Osia insertion proceeded after flap elevation and haemostasis to avoid monopolar diathermy use once the implant was in place. The skin was retracted and the dissection extended posteriorly, with deep dissection into the subperiosteal plane to expose the site for BI300 screw placement. The periosteum was retracted superiorly and anteriorly, allowing subperiosteal placement of the magnet-bearing component and transducer. This technique aimed to maximise soft tissue coverage over the implant in the context of reduced skin thickness following TPF harvest. Standard layered closure was performed. Skin grafts were inset over the reconstructed ear, and a protective silicone cup-like dressing was applied over the ear, secured by a crepe bandage. This was removed at one week follow-up.

In the case of Osia implantation without concurrent reconstruction, a lazy-J incision was made to the subcutaneous plane, followed by a staggered posterior incision down to the subperiosteal plane to preserve vascular supply for potential future microtia reconstruction. In another case where microtia reconstruction had already been completed, a posteriorly based horizontal incision was made across the waist of the Osia to avoid compromising vascular supply.

In all cases, a 15 mm skin margin was preserved between the incision and edge of the implant. The 3 mm BI300 screw was used universally due to limited paediatric bone thickness and lack of added benefit from a 4 mm screw in an active implant. In revision cases involving conversion from a Baha Attract to Osia, the existing BI300 screw was used to

avoid dural injury with attempted screw removal.

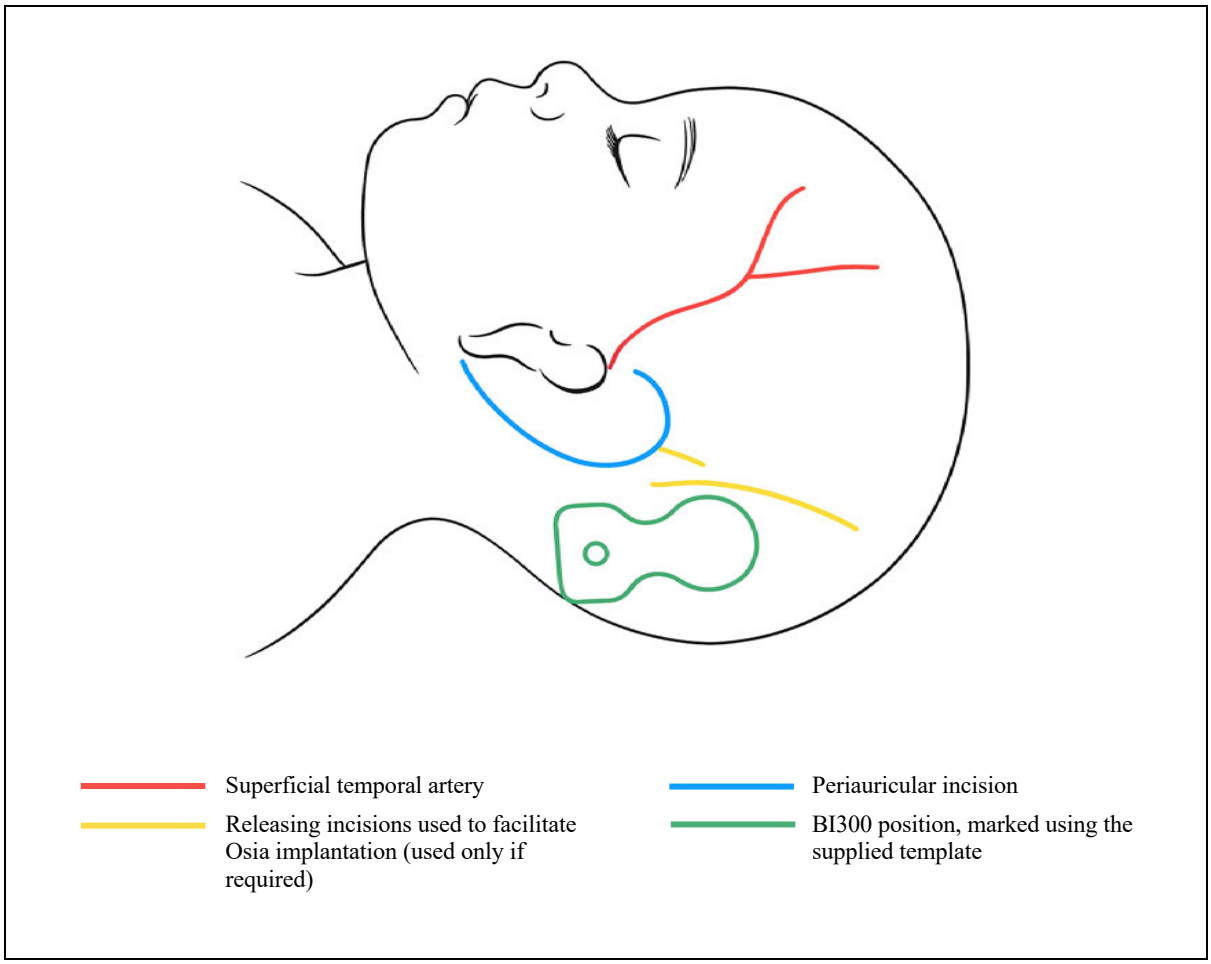

FIGURE 2. Surgical Markings for Combined Osia Implantation with Auricular Reconstruction.

Data collection

Clinical records, operative reports, and audiological assessments were reviewed. Data collected included demographic details and clinical history (laterality of microtia-atresia, presence of syndromes or comorbidities, prior hearing devices, and duration of follow-up). Skull thickness was measured on axial temporal bone computed tomography (CT), defined as the mean of the thickest and thinnest points in the retrosigmoid region at the level of the lateral semicircular canal. Surgical data included concurrent auricular reconstruction or

device conversion, intra-operative complications, post-operative complications, and length of hospital stay.

Pre-operative thresholds were obtained as ear-specific air conduction (AC) and bone conduction (BC) audiometry under headphones or insert earphones. Post-operative outcomes were assessed using Osia-aided soundfield thresholds, with contralateral masking. In cases where soundfield data were unavailable, BC Direct thresholds from the Cochlear™ fitting software were recorded. To account for potential differences between BC Direct and soundfield thresholds, sensitivity analysis was also performed. All PTAs were calculated across 0.5, 1, 2, and 4 kHz thresholds.

As unaided soundfield thresholds were not routinely collected, conventional functional gain could not be calculated. Instead, two complementary outcome measures were derived:

1. AC-to-aided improvement – the difference between pre-operative AC PTA and Osia-aided PTA, reflecting the degree of correction of the conductive component, though not equivalent to functional gain.
2. Effective gain – the difference between Osia-aided PTA and pre-operative BC PTA, demonstrating the extent to which aided thresholds approximated the cochlear reserve (94).

Speech audiology was performed variably across centres using a variety of age-appropriate measures. These included the Speech Perception in Noise (SPIN) sentence test, which assesses recognition of everyday sentences in background noise, and the Bamford-Kowal-Bench (BKB) test, a validated measure of sentence recognition for children administered in quiet or noise.

Parent-reported outcomes (PROMs) were assessed using the Parents' Evaluation of Aural/Oral Performance of Children (PEACH) questionnaire, a validated tool developed to evaluate the effectiveness of hearing devices based on parents' observations, suitable from infancy through to school age (95). Device use was collected as part of the PEACH questionnaire on a 5-point ordinal scale (0 = Never, 1 = Rarely, 2 = Sometimes, 3 = Often, 4 = Always). Additional qualitative feedback was extracted from unstructured parental comments recorded at follow-up.

Statistical analysis

Statistical analysis was conducted with SPSS statistics software, version 29.0 (IBM Corp., Armonk, NY, USA). Continuous variables are reported as median [interquartile range]. Given the small sample size, statistical analyses were performed to provide descriptive insight rather than to test hypotheses. Non-parametric tests (Wilcoxon signed-rank) were used for pre–post comparisons, but these were treated as exploratory and intended only to illustrate the direction and consistency of changes rather than to infer population-level effects.

Results

Patient characteristics

Nine children with microtia-atresia underwent implantation of 10 Osia devices between 2022 and 2025 and met inclusion criteria (Table 1). The median age at implantation was 8.6 years [IQR 5.4-11.6; range 4.8-12.8]. Six implants were performed in males and four in females. Five children (56%) had associated hemifacial microsomia without any other congenital syndrome. The implanted side was right in five children, left in three children and bilateral in one child, who underwent sequential implantation at six and eight years of age. All but one child had prior experience with passive bone conduction devices: five with softbands and

three with ipsilateral Baha Attract systems. The three patients with Baha Attracts underwent concurrent conversion from Baha Attract to Osia. The majority of implants (n = 8) were performed concurrently with microtia reconstruction, one was staged after reconstruction (which had been undertaken overseas), and one implant was undertaken as a standalone procedure without auricular reconstruction. Median follow-up duration was 13.0 months [3.8-25].

TABLE 1. Patient demographics and clinical characteristics

Patient ID	Age at surgery (yrs)	Sex	Side implanted	Comorbidities	Previous device	Follow-up duration (mo)
1	5.0	Male	Right	Hemifacial microsomia	Baha softband	4
2	4.8	Male	Left	-	Baha softband	36
3 (L)	6.6	Female	Left	Hemifacial microsomia	Baha softband	34
3 (R)	8.7	Female	Right	Hemifacial microsomia	Baha softband	8
4	9.5	Male	Right	-	Baha Attract	14
5	5.5	Female	Right	Hemifacial microsomia	Baha softband	12
6	12.6	Male	Right	-	Baha Attract	17
7	11.2	Female	Left	Hemifacial microsomia	None	3
8	8.5	Male	Right	-	Baha Attract	3
9	12.8	Male	Left	Hemifacial microsomia	Baha softband	22

Surgical outcomes

Surgical characteristics and outcomes are summarised in Table 2. Median bone and soft tissue thickness on pre-operative CT PTB was 4 mm [2.7-5.45] and 3.7 mm [3.1-6.95] respectively.

One patient had intraoperative bleeding from a prominent emissary vein which was controlled intraoperatively with bone wax without further sequelae. Dural exposure was noted in seven cases, none of which were associated with dural injury or CSF leak. Bone polishing was not required in any cases. All combined cases stayed overnight and were discharged home the following day. Of the two standalone implant cases, one patient stayed overnight due to interstate travel, while the other was managed as a day procedure.

TABLE 2. Surgical characteristics and outcomes

Patient ID	Implant laterality	Additional procedures	Intra-op complications	Post-op complications	Length of stay (nights)
1	Right	-	-	-	0
2	Left	Microtia reconstruction	-	-	1
3 (L)	Left	Microtia reconstruction	Emissary vein bleed (controlled)	Erythema (resolved)	1
3 (R)	Right	-	-	Wound infection (resolved)	1
4	Right	Conversion from Baha Attract + microtia reconstruction	-	-	1
5	Right	Microtia reconstruction	-	-	1
6	Right	Conversion from Baha Attract + microtia reconstruction	-	-	1
7	Left	Microtia reconstruction	-	Non-healing pressure sore → explant	1
8	Right	Conversion from Baha Attract + microtia reconstruction	-	-	1
9	Left	Microtia reconstruction	-	Insect bite → skin breakdown → explant	1

Two minor post-operative complications (Clavien-Dindo Grade I) were reported. The first involved mild erythema at the magnet site shortly after switch-on, consistent with transient soft tissue irritation. This resolved spontaneously with temporary discontinuation of device wear and required no medical intervention. The second minor event was a superficial wound infection at the incision site three weeks post-operatively, presenting with localised erythema, which settled promptly with a short course of oral antibiotics.

Two major complications (Clavien-Dindo Grade IIIb) were reported and required device explantation. In the first case, a focal non-healing pressure area developed directly over the magnet site around the edge of the flap beneath the edge of the cup-like dressing. Despite early off-loading measures, the ulcer progressed to full-thickness soft-tissue breakdown with exposure of the magnet, requiring explantation. The second major complication reportedly arose following an insect bite at the posterior edge of the post-auricular incision. This resulted in progressive skin breakdown with localised cellulitis. Delayed presentation for medical review limited opportunities for early intervention, and subsequent flap revision failed to achieve durable tissue coverage, again necessitating device removal.

Audiological outcomes

Pre-operative audiometry demonstrated near-normal cochlear reserve across the cohort, with a median BC PTA of 8.1 dB HL [3.8-10.9] and a median air-bone gap of 58.8 dB [53.1-62.5]. Following implantation, Osia-aided thresholds showed substantial improvement, with a median aided PTA of 16.3 dB HL [13.8-20.0]. The median AC-to-aided improvement was 52.5 dB [33.8-56.3], reflecting effective correction of the conductive component. Effective gain, defined as Osia-aided PTA relative to BC PTA, demonstrated that aided thresholds closely approximated the cochlear reserve (median 10.0 dB HL; IQR: 5.0-20.0). A sensitivity analysis restricted to patients with aided soundfield data produced comparable results, with a median aided PTA of 20 dB HL [15.6-20.6] and effective gain of 20.0 dB [12.5-20.6] (Table 3).

One patient (ID 6) recorded an implausible Osia-aided PTA that exceeded the pre-operative BC PTA. This discrepancy may reflect overestimation of BC PTA due to variability in

paediatric BC audiometry, together with underestimation from BC Direct calibration. This patient was therefore excluded from pooled analyses.

Speech audiometry was performed variably across centres but consistently demonstrated postoperative improvement. Osia-aided BKB and SPIN sentence scores of 90-100% were observed with four implants, with one showing improvement in SPIN from 48% unaided to 90% Osia-aided.

TABLE 3. Audiological outcomes

	Unaided			***Osia-aided***			
Patient ID	BC PTA (dB HL)	AC PTA (dB HL)	ABG (dB)	SF PTA (dB HL)	AC-to-aided improvement (dB HL)	EG (dB)	Speech outcomes
1	8.8	71.3	62.5	13.8^	52.5	10.0	-
2	10.0	68.8	58.8	8.8^	55.0	3.8	-
3 (L)	0.0	53.8	53.8	20.0	33.8	20.0	PLOTT 7/7 SRT -2.1 BKB 100%
3 (R)	0.0	51.3	51.3	21.3	30.0	21.3	BKB 90%
4	10.0	72.5	62.5	11.3^	56.3	6.3	-
5	5.0	63.8	58.8	11.3^	47.5	11.3	BKB 100%
6	21.3	82.5	61.3	1.3^	76.3	-15.0+	SPIN sentences 90% (vs 48% unaided)
7*	13.8	63.8	50.0	-	-	-	-
8	6.3	70.0	63.8	11.3	58.8	5.0	-
9*	7.5	61.3	53.8	-	-	-	-

Abbreviations: AC = air conduction; ABG = air-bone gap; BC = bone conduction; BKB = Bamford-Kowal-Bench sentence test; dB = decibels; EG = effective gain; HL = hearing level; PLOTT = Phoneme Detection and Imitation Test; PTA = pure tone average; SF = soundfield; SPIN = Speech Perception in Noise test; SRT = speech reception threshold

Notes:
* = Device explanted prior to switch on
+ = implausible value; patient excluded from analysis
^ = PTA calculated from BC Direct rather than soundfield thresholds

Patient-Reported Outcomes

Pre- and post-operative PEACH scores were available for seven implants (Table 4). In quiet, median scores improved from 85% [70-85] pre-operatively with previous devices to 90% [90-100] with Osia-amplification. In noise, scores improved from 65% [50-80] to 80% [75-85]. Global PEACH scores increased by +10 [7.8-22.5], with all children showing gains. Median device use scores increased from 2 [1-2.5] ("Rarely/Sometimes") to 4 [3.5-4] ("Often/Always"). Non-parametric comparisons (Wilcoxon signed-rank) suggested statistically significant improvements (p <0.05 for all comparisons); however these findings should be interpreted cautiously given the very small sample size and the descriptive intent of the analysis.

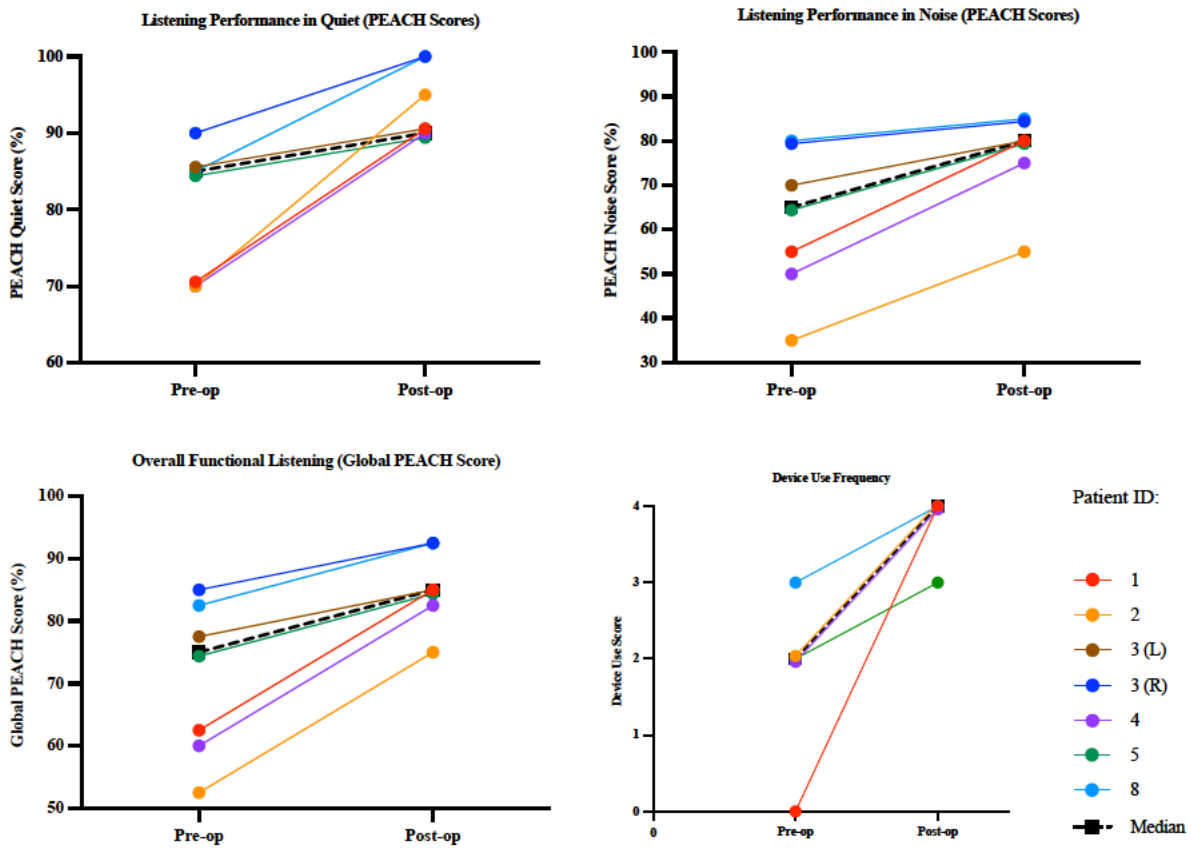

FIGURE 3. Functional Hearing Outcomes Before and After Osia Implantation

Qualitative feedback reinforced the quantitative outcomes (Table 5). Parents consistently reported substantial improvements in listening and daily functioning with Osia compared to previous devices. Families noted markedly increased compliance, with children who previously wore softband or Baha attract devices inconsistently now using the Osia system from waking until bedtime. Parents described reductions in listening fatigue, better classroom participation, and noticeable behavioural improvements.

Practical benefits were frequently highlighted, with the Osia regarded as easier to manage and more comfortable than softband devices, which had often caused sensory discomfort and fitting challenges. One child (Patient 3) experienced listening difficulties following staged bilateral implantation. After her first implant, she reported improved sound quality and consistent Osia use. However, following contralateral implantation two years later, she experienced excessive loudness, echoing, and a preference for unilateral use. With reprogramming, her tolerance improved, and over time she benefitted from bilateral input. This was reflected in her 12-month PEACH score after bilateral implantation, which exceeded the 12-month PEACH score following the first implant alone.

TABLE 4. Thematic Summary of Parental Feedback

Theme	Representative Feedback
Compliance & daily use	"Now wears [the Osia] all day; previously resisted softband" "Never wore attract, now enjoys wearing [the Osia] daily" "Wearing [the Osia] every day since switch on" "Dislikes it when [the Osia] goes for servicing"
Listening fatigue	"Noticeably less tired" "Improved listening and behaviour"
Communication & social participation	"More confident and content" "Better speech and connecting better with peers"
Comfort & usability	"No longer complains of discomfort" "Easy to use" "Doesn't even feel [the Osia]" "Taking responsibility for looking after [the Osia]"

Discussion

This case series demonstrates that Osia implantation in children 12 years and under is feasible, with consistent audiological benefit and high parental satisfaction. Notably, most procedures were performed in conjunction with microtia reconstruction using a 3D-printed porous polyethylene framework, representing a novel combined approach in the Australian setting.

Several active BCIs are now available, each with distinct device profiles, advantages and limitations in paediatric practice (Figure 2, Table 5). Among these, the Osia has the widest fitting range (up to 55 dB HL), compared with 45 dB HL for other available devices. Unlike electromagnetic systems such as the BONEBRIDGE® (MED-EL®, Innsbruck, Austria) and Sentio™ (Oticon Medical™, Askim, Sweden), the Osia provides conditional 3T MRI compatibility with a smaller artefact, which can be further reduced with surgical removal of the magnet cassette - an important consideration in children with complex comorbidities who may require repeated imaging (Table 5).

Notably, it is the only atBCI that does not require drilling of a bony well – an advantage in younger patients with thinner skulls. However, because the device rests on the bone surface rather than recessed within it, pressure-related soft tissue complications may occur, particularly in microtia patients where fascial rotation flaps reduce tissue thickness over the device, as observed in two cases in this series. This balance between surgical simplicity and soft tissue stress must be considered when selecting candidates and surgical devices, especially in the context of combined auricular reconstruction.

TABLE 5. Comparison of Active Transcutaneous Bone Conduction Implants

Feature	MED-EL BONEBRIDGE (BCI 602)	Cochlear Osia (OSI300)	Oticon Sentio (Ti implant)
Transducer thickness (mm)	8.6	4.9	5
Magnet thickness (mm)	4.4	3.9	4
Bone bed preparation	3.5-4.5 mm well	-	3 mm well
Fixation screw	2x 5 mm screws	1x 3.2 or 4.2 mm screw	2x 2.7 mm screws
Bone removal volume	~910 mm^3 (with 1 mm lift), ~1170 mm^3 (without lifts) + volume from 2x screws	70 mm^3 (for 4.2 mm screw)	~400-700 mm^3 + volume from 2x screws
Transducer Type	Electromagnetic	Piezoelectric (ceramic)	Electromagnetic
MRI Compatibility	1.5T conditional	1.5T and 3T conditional	1.5T conditional
MRI Artefact	Up to 15 cm	7.5-11.6 cm (with magnet); 2.8-6.0 cm (without magnet) (at 3T)	Approximately 18 cm
Fitting range	Up to 45 dB HL	Up to 55 dB HL	Up to 45 dB HL
TGA approved age range (date of approval)	≥5 years (May 2022)	≥5 years (March 2024)	≥12 years (May 2025)
FDA approved age range (date of approval)	≥12 years (Sept 2019)	≥5 years (April 2024)	≥12 years (July 2024)

dB HL = decibels hearing level; FDA = United States Food and Drug Administration; MRI = Magnetic Resonance Imaging; T = Tesla; TGA = Australian Therapeutic Goods Administration

In this case series, intra-operative complications were rare, with only one minor bleeding episode. Two minor post-operative complications arose which were managed conservatively. However, two major post-operative complications led to device explantation. Both complications unexpectedly occurred in older children (aged 11 and 12 at the time of surgery respectively), although each had substantial hemifacial microsomia. In these patients, thin soft tissue and wound tension likely contributed to skin breakdown. In one child, the complication occurred under unusual circumstances, precipitated by an insect bite overlying the bulk of the piezoelectric transducer, an area already under tension. In the second, skin breakdown occurred at the superior edge of the device, adjacent to the silicone housing the magnet (the thinnest part of the device), prior to switch on. This was likely due to thinning of the overlying soft tissue with flap harvest and, post-operatively, localised pressure from the cup dressing. The patient was fitted with a softband, with revision surgery planned after 12 months to allow for tissue growth and revascularisation. Additional challenges in both cases

included delay in seeking wound care and long-distance follow-up, which complicated wound surveillance and early intervention.

Most Osia complications described in the paediatric literature are minor and conservatively managed, including skin irritation, erythema, seromas, mild swelling and infection, pain and headaches. Published complication rates for Osia devices in children range from 0 to 34%, with major complication rates under 4% (66-77). However, these figures are largely drawn from small observational and pilot studies, most of which do not focus on children with complex craniofacial malformations or combined reconstructive procedures. The higher rate observed in our series likely reflects both the small cohort size, where single events disproportionately affect percentages, and the increased surgical complexity of patients with hemifacial microsomia undergoing combined reconstructive procedures. Collectively, these findings highlight that while Osia implantation is technically safe, careful post-operative surveillance and meticulous wound management are critical, particularly in children with craniofacial anomalies undergoing combined reconstructive procedures.

Novel surgical techniques may play a role in improving outcomes in anatomically complex cases. For example, partial recessing of the Osia may help reduce pressure on the overlying skin. This could make the Osia a unique compromise between conventional Osia implantation and the BONEBRIDGE. Unlike the BONEBRIDGE, which requires recessing in a bony well, the Osia does not – but emerging techniques suggest it could be recessed when advantageous. Bere et al described a minimally invasive subperiosteal pocket technique for Osia implantation without bone fixation, with early results suggesting that omitting fixation does not compromise device function (96). Similarly, Alnoury et al reported an endoscope-assisted minimally invasive approach that allows precise screw alignment through

a small incision (73). These innovations, by reducing incision size, minimising drilling in thin skulls, and potentially enabling recessed placement of the Osia without reliance on bone thickness for screw fixation, may lower the risk of soft tissue complications while maintaining device performance.

In terms of audiological benefit, the Osia demonstrated aided thresholds closely approximating cochlear reserve across the cohort. Although conventional functional gain could not be calculated, effective gain served as a clinically meaningful proxy and confirmed the ability of the Osia to restore hearing sensitivity to near-normal levels. These findings are consistent with prior studies reporting mean functional gains of 43–45 dB with Osia (44, 66, 67, 72). Speech perception outcomes, though assessed variably across centres, similarly indicated excellent Osia-aided performance. Challenges in obtaining speech measures in some children, particularly due to inattentiveness, underscored the practical difficulties of formal testing in this population. PROMs, assessed using the PEACH questionnaire, confirmed meaningful real-world benefits, including improved listening in quiet and in noise, classroom participation, and social confidence. These outcomes mirror the broader literature and highlight the importance of PROMs in young children where formal audiological assessment may be challenging (66, 68, 73, 76).

Only one child in this series, who underwent staged bilateral implantation, faced listening difficulties after implantation of the second side two years after the first. These challenges improved with reprogramming and persistent device use, with improved PEACH scores at 12-month follow-up. A similar phenomenon was reported by Gordon et al. in two children with prior percutaneous bone conduction on one side later receiving an Osia on the contralateral side (76). Both initially expressed a preference for their first ear despite

demonstrating excellent speech perception with the Osia, but improved with counselling and reprogramming. These findings suggest that auditory adaptation may be important in sequential bilateral implantation to support successful long-term use. Further investigation is warranted to determine whether simultaneous bilateral implantation is better tolerated in this population.

Importantly, patients who converted from passive transcutaneous or softband devices demonstrated high daily use with the Osia, in contrast to the reported minimal use of their previous devices, and expressed a strong preference for the new device – a sentiment echoed in the literature (66, 72). This highlights a critical but under-recognised issue: while clinicians may assume that children with unilateral conductive hearing loss are adequately rehabilitated with non-surgical or passive devices, actual device use is often limited. Particularly in children, device acceptability and comfort drive real-world effectiveness as much as audiological gain. Evidence from the cochlear implant literature underscores the developmental importance of providing consistent auditory input during sensitive periods of language and social development (3, 16-26, 29, 31-33, 97-114). Poor compliance risks leaving children with unaddressed auditory deprivation, with potential long-term consequences for neuroplasticity, language development, and social participation.

Many contemporary hearing devices, including BAHA Attract and Osia, incorporate automatic data logging functions. Although limited studies have reported objective usage data, available evidence suggests excellent compliance with Osia, with mean or median daily wear times of 9.8-10.3 hours at 6 months post-operatively (68, 70). Future studies incorporating device-logged data will be valuable in quantifying paediatric compliance with passive versus active bone conduction systems. Such information could help clarify the true

burden of unaddressed hearing loss in these children and guide evidence-based decisions about the most effective rehabilitative strategies and timing for surgical intervention.

The main limitation of this study is the small sample size. This reflects both the rarity of microtia-atresia and combined reconstructive surgery, as well as the recent regulatory approval of Osia implantation in younger children in Australia, where implantation under 12 years previously required special authorisation. Although improvements in PTA and PEACH scores reached statistical significance, interpretation is limited by the small sample size, and these results should be viewed as hypothesis-generating rather than definitive. The retrospective design introduces additional constraints, including inconsistency in outcome reporting and missing data for some patients. At the time of this study, few centres in Australia were performing alloplastic microtia reconstruction, resulting in many patients travelling from interstate, and one from overseas. As a result, post-operative auditory rehabilitation was delegated to audiologists local to patients, introducing variability in testing and outcome reporting. Follow-up duration was relatively short, preventing assessment of long-term device survival, complications and functional outcomes.

Despite these limitations, the series contributes the first Australian data on paediatric Osia outcomes and one of the earliest reports internationally of combined Osia implantation with single-stage alloplastic auricular reconstruction. It highlights practical considerations for combined reconstructive approaches and underscores concerns that passive devices are not necessarily providing adequate auditory rehabilitation due to poor compliance - an issue not always apparent on routine clinical assessment and audiometric testing. Looking ahead, multicentre collaboration with standardised outcome reporting and long-term follow-up will

aid in building evidence in this patient population, particularly among patients undergoing concurrent atBCI implantation and microtia reconstruction.

Conclusion

Osia implantation in children with microtia-atresia aged 12 years and younger is feasible and provides consistent audiological and functional benefit. When combined with auricular reconstruction, it offers an opportunity for comprehensive early rehabilitation of both form and function. Careful surgical planning and close post-operative surveillance are essential to mitigate wound complications, particularly in children with craniofacial malformations undergoing combined surgery. Early findings highlight the importance of considering device comfort and compliance alongside functional gain. Larger multicentre studies with long-term follow-up are required to clarify long-term safety, durability and developmental impact.

Chapter 4: Conclusions and Future Directions

Summary of Findings

This thesis examined the outcomes of children with microtia-atresia, with a particular focus on hearing rehabilitation using active transcutaneous bone conduction implants (atBCIs), specifically the Osia® system (Cochlear™ BAS, Gothenburg, Sweden). The work comprised two main components: first, a systematic review of the literature evaluating the surgical, audiometric and patient-reported outcomes of paediatric Osia implantation, and second, a retrospective case series reporting early Australian experience with Osia implantation in children 12 years and under with microtia-atresia, the majority of whom underwent concurrent alloplastic auricular reconstruction.

From the systematic review presented in Chapter 2, early paediatric evidence for the Osia indicates consistent audiological benefit and a favourable safety profile, with functional gains and low complication rates comparable to those reported in adult populations. In the case series presented in Chapter 3, paediatric Osia implantation was found to be feasible and safe in carefully selected patients, again demonstrating consistent functional gains and improvements in patient-reported outcome measures. Notably, concurrent single-stage alloplastic reconstruction was shown to be technically feasible, and offers the advantage of reducing repeated surgeries, anaesthetic exposure and cumulative hospitalisations for select children and their families. However, the occurrence of complications in two children with craniofacial anomalies undergoing combined procedures highlights the need for careful patient selection and vigilant follow-up to ensure soft tissue durability.

Limitations

The limitations of this thesis largely reflect the constraints inherent in both the existing literature and the case series' design. Small cohort sizes, though expected given the rarity of microtia-atresia and the recent regulatory approval for Osia implantation in younger children, limit statistical power and reduce generalisability. Short follow-up durations in both the systematic review and case series prevented assessment of long-term complications, device reliability and durability of functional listening improvements over time.

In the systematic review, heterogeneity in study design, outcome measures, and reporting formats hindered direct comparisons and meta-analysis, underscoring the need for greater standardisation in paediatric hearing research. In the case series, the retrospective design introduced reliance on the availability and completeness of clinical records, with potential for missing data and selection bias. The inherent challenges of performing detailed audiological and speech testing in young children – particularly issues of attention and test reliability – further constrained the dataset.

Clinical Implications

The findings of this thesis suggest that the Osia is a viable, safe, and well-tolerated option for hearing rehabilitation in younger children with microtia-atresia, providing measurable audiological gains and functional benefits that are highly valued by patients and families. In clinical practice, device selection and timing of implantation should be tailored to the individual child's anatomy, age, developmental profile, and family preferences. Particular consideration should be given to children with craniofacial comorbidities, where outcomes may differ, and to those undergoing concurrent auricular reconstruction, where reduced soft tissue thickness or vascularity may impact both short- and long-term outcomes.

Multidisciplinary planning between the otologist, reconstructive surgeon and audiologist is likely to optimise results in this subgroup.

Importantly, the evidence to date – including the data presented in this thesis – suggests that children with unilateral hearing loss derive substantial benefit from Osia implantation, despite the historical tendency to underestimate the functional impact of unilateral CHL. Incorporating validated, child-reported outcome measures into routine practice can help capture these real-world benefits and should support counselling of families and shared decision making. As the evidence base expands and atBCI technologies continue to evolve, there will be increasing scope to establish evidence-based guidelines for the management of children with microtia-atresia. Such guidelines will be critical to ensuring equitable and consistent access to interventions that may improve communication, classroom participation and long-term developmental outcomes.

Future Directions

Future research in paediatric microtia-atresia should prioritise prospective, multicentre studies to build larger cohorts and enable meaningful subgroup analyses stratified by age, device type and comorbidities. Standardisation of outcome measures is essential, with consistent use of validated, paediatric-specific tools for speech perception, spatial hearing, functional listening, and quality of life, allowing results to be compared across centres and pooled over time. Comparative studies of the Osia system alongside other atBCIs such as the BONEBRIDGE® (MED-EL®, Innsbruck, Austria) and Sentio™ (Oticon Medical™, Askim, Sweden) would provide valuable information on relative performance, user comfort, complication rates, and cost-effectiveness in paediatric populations. International

collaboration and the establishment of registries would further support longitudinal data collection, monitoring of device reliability and benchmarking across centres.

Particular attention should be directed to children with craniofacial and ENT-related comorbidities, who comprise a sizeable proportion of those receiving atBCIs. These children may present unique anatomical and physiological challenges, including reduced bone volume, altered vascularisation, or syndromic neurodevelopmental differences that could affect both device performance and real-world listening outcomes. Likewise, the influence of auricular reconstruction – whether prior, simultaneous, or staged – warrants systematic evaluation. In particular, the long-term durability of overlying soft tissue and risk of skin or flap complications in reconstructed ears remain insufficiently understood and could have implications for device longevity and revision rates.

Further work is also needed to address the ongoing uncertainty around the management of unilateral conductive hearing loss (CHL) in microtia-atresia. Despite growing evidence that unilateral CHL can have significant functional, educational, and psychosocial consequences as outlined in Chapter 1, consensus on the timing and indications for amplification remains lacking. Importantly, studies suggest that children consistently rate their own listening difficulties as greater than those perceived by parents, highlighting the risk of underestimating the burden of unilateral CHL (34, 76). Prospective research is needed to quantify functional performance, listening fatigue, and academic outcomes in this group. In particular, defining the optimal timing of implantation – balancing the technical considerations against the potential developmental and psychosocial benefits of aiding children prior to school entry – will be crucial. Ultimately, such work could inform the

development of guidelines on aiding children with unilateral microtia-atresia and other causes of unilateral hearing loss, an area where practice remains variable and often anecdote-driven.

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Appendix 1: Search Strategy

OVID MEDLINE

1. Cochlear* adj2 Osia.mp.
2. Osia.mp.
3. Bone-Anchored Prosthesis.mp.
4. cochlear*.mp.
5. ear*.mp.
6. hear*.mp.
7. 4 or 5 or 6
8. 3 and 7
9. 1 or 2 or 8
10. limit 9 to humans

SCOPUS

((TITLE-ABS-KEY (osseointegrated) AND TITLE-ABS-KEY (cochlear AND implant*)))

OR (TITLE-ABS-KEY (osia))

LIMIT TO humans

EMBASE

1. Cochlear* adj2 Osia.mp.
2. Osia.mp.
3. Bone-Anchored Prosthesis.mp.
4. cochlear*.mp.
5. ear*.mp.
6. hear*.mp.
7. 4 or 5 or 6
8. 3 and 7
9. 1 or 2 or 8
10. limit 9 to humans

Appendix 2: Risk of Bias Assessments

JBI Critical Appraisal Checklist for Case Series	
Citation: Mozaffari, M., Guderley, N., Wong, A., Konstantinidou, S., Amin, N., & Nash, R. (2025). Surgical and audiometric outcomes of active osseointegrated bone-conduction hearing device (Cochlear TM Osia R 2 system) placement from a tertiary paediatric centre. International Journal of Pediatric Otorhinolaryngology, 190, 112272. doi:https://dx.doi.org/10.1016/j.ijporl.2025.112272	
Checklist item	**Appraisal**
1. Were there clear criteria for inclusion in the case series?	Yes
2. Was the condition measured in a standard, reliable way for all participants included in the case series?	Yes
3. Were valid methods used for identification of the condition for all participants included in the case series?	Yes
4. Did the case series have consecutive inclusion of participants?	Yes
5. Did the case series have complete inclusion of participants?	Yes
6. Was there clear reporting of the demographics of the participants in the study?	Yes
7. Was there clear reporting of clinical information of the participants?	Yes
8. Were the outcomes or follow-up results of cases clearly reported?	Yes
9. Was there clear reporting of the presenting site(s)/clinic(s) demographic information?	Yes
10. Was statistical analysis appropriate?	Yes
Overall risk of bias	Low
• **Strengths:** consecutive inclusion, comprehensive reporting, adequate outcome measures, clear surgical and audiological methods. • **Limitations:** retrospective design, lack of standardised PROMs, informal reporting of compliance, no objective device usage data (acknowledged by authors).	

JBI Critical Appraisal Checklist for Case Series	
Citation: You, P., Choi, A., Drob, J., Hunsaker, S. M., Liu, Y. C., & Silva, R. (2022). Early Outcomes of a New Active Transcutaneous Bone Conduction Implant in Pediatric Patients. Otology & Neurotology, 43(2), 212-218. doi:https://dx.doi.org/10.1097/MAO.0000000000003426	
Checklist item	**Appraisal**
1. Were there clear criteria for inclusion in the case series?	Yes
2. Was the condition measured in a standard, reliable way for all participants included in the case series?	Yes
3. Were valid methods used for identification of the condition for all participants included in the case series?	Yes
4. Did the case series have consecutive inclusion of participants?	Unclear
5. Did the case series have complete inclusion of participants?	No
6. Was there clear reporting of the demographics of the participants in the study?	Yes
7. Was there clear reporting of clinical information of the participants?	Yes
8. Were the outcomes or follow-up results of cases clearly reported?	Yes
9. Was there clear reporting of the presenting site(s)/clinic(s) demographic information?	Yes
10. Was statistical analysis appropriate?	Yes
Overall risk of bias	Moderate
• **Strengths:** clear inclusion/exclusion, standardised audiometry, detailed reporting of demographics/outcomes. • **Limitations:** exclusion of patients without aided testing introduces potential selection bias; relatively short mean follow-up (7 months); retrospective design.	

JBI Critical Appraisal Checklist for Case Series
Citation: Florentine, M. M., Virbalas, J., & Chan, D. K. (2022). Early surgical and audiologic outcomes of active, transcutaneous, osseointegrated bone-conduction hearing device (Osia 2 R system) placement.

International Journal of Pediatric Otorhinolaryngology, 156, 111114. doi:https://dx.doi.org/10.1016/j.ijporl.2022.111114	
Checklist item	**Appraisal**
1. Were there clear criteria for inclusion in the case series?	Yes
2. Was the condition measured in a standard, reliable way for all participants included in the case series?	Yes
3. Were valid methods used for identification of the condition for all participants included in the case series?	Yes
4. Did the case series have consecutive inclusion of participants?	Yes
5. Did the case series have complete inclusion of participants?	Yes
6. Was there clear reporting of the demographics of the participants in the study?	Yes
7. Was there clear reporting of clinical information of the participants?	Yes
8. Were the outcomes or follow-up results of cases clearly reported?	Yes
9. Was there clear reporting of the presenting site(s)/clinic(s) demographic information?	Yes
10. Was statistical analysis appropriate?	Yes
Overall risk of bias	Low
• **Strengths:** consecutive inclusion, complete reporting of demographics, surgical and audiological outcomes, standardised measurement. • **Limitations:** relatively short mean follow-up (23 weeks), no within-subject statistical comparisons of pre- vs post-op aided thresholds.	

JBI Critical Appraisal Checklist for Case Series	
Citation: Alnoury, M. K., & Daniel, S. J. (2024). Minimally Invasive OSIA Bone Conduction Hearing Implant (MOSIA) in Children: How I do it? Laryngoscope, 134(4), 1901-1906. doi:https://dx.doi.org/10.1002/lary.31001	
Checklist item	**Appraisal**
1. Were there clear criteria for inclusion in the case series?	Yes
2. Was the condition measured in a standard, reliable way for all participants included in the case series?	Yes
3. Were valid methods used for identification of the condition for all participants included in the case series?	Yes
4. Did the case series have consecutive inclusion of participants?	Unclear
5. Did the case series have complete inclusion of participants?	Yes
6. Was there clear reporting of the demographics of the participants in the study?	Some
7. Was there clear reporting of clinical information of the participants?	Yes
8. Were the outcomes or follow-up results of cases clearly reported?	Yes
9. Was there clear reporting of the presenting site(s)/clinic(s) demographic information?	No
10. Was statistical analysis appropriate?	N/A
Overall risk of bias	Moderate
• Strengths: clear inclusion • Limitations: small sample size (n=7), short follow-up, unclear consecutive recruitment, limited demographics, site not specified.	

JBI Critical Appraisal Checklist for Case Series	
Citation: Stevens, S. M., Meyer, A., Rivas, A., Mowry, S., Carvalho, D., Chang, K. W., . . . Tejani, V. (2025). Outcomes Following Cochlear Osia 2 Implantation in Patients Ages 5-11 Years: A Multi-Center Trial. Laryngoscope, 135(8), 2958-2966. doi:10.1002/lary.32159	
Checklist item	**Appraisal**
1. Were there clear criteria for inclusion in the case series?	Yes
2. Was the condition measured in a standard, reliable way for all participants included in the case series?	Yes
3. Were valid methods used for identification of the condition for all participants included in the case series?	Yes
4. Did the case series have consecutive inclusion of participants?	Yes
5. Did the case series have complete inclusion of participants?	1 incomplete
6. Was there clear reporting of the demographics of the participants in the study?	Yes

7. Was there clear reporting of clinical information of the participants?	Yes
8. Were the outcomes or follow-up results of cases clearly reported?	Yes
9. Was there clear reporting of the presenting site(s)/clinic(s) demographic information?	Yes
10. Was statistical analysis appropriate?	Yes
Overall risk of bias	Low
• **Strengths**: prospective consecutive enrolment, standardised outcome measures, multicentre design, low attrition rate. • **Limitations**: incomplete follow-up for 1 patient not fully explained.	

JBI Critical Appraisal Checklist for Cohort Studies	
Citation: Zawawi, F., Garrada, M., & Bukhari, A. F. (2025). Exploring Osia System's auditory outcome and the surgical benefits of posterior placement of BI300 in children with microtia. International Journal of Pediatric Otorhinolaryngology, 195, 112398. doi:https://doi.org/10.1016/j.ijporl.2025.112398	
Checklist item	**Appraisal**
1. Were the two groups similar and recruited from the same population?	Yes
2. Were the exposures measured similarly to assign people to both exposed and unexposed groups?	Yes
3. Was the exposure measured in a valid and reliable way?	Yes
4. Were confounding factors identified?	Partially – concurrent reconstruction/syndromes reported; previous reconstruction/microtia severity could also influence outcomes
5. Were strategies to deal with confounding factors stated?	Limited – group allocation based on reconstruction feasibility which introduced potential bias. No adjustment reported.
6. Were the groups/participants free of the outcome at the start of the study (or at the moment of exposure)?	Yes
7. Were the outcomes measured in a valid and reliable way?	Yes
8. Was the follow-up time reported and sufficient to be long enough for outcomes to occur?	Yes
9. Was follow-up complete, and if not, were the reasons to loss to follow-up described and explored?	Yes
10. Were strategies to address incomplete follow-up utilized?	No loss to f/u reported
11. Was appropriate statistical analysis used?	Yes
Overall risk of bias	Low to moderate
• **Strengths**: Prospective design, full cohort inclusion, detailed demographics and outcomes, appropriate non-parametric statistics. • **Limitations**: Small sample size (n=23), single centre, no adjusting for confounders, potential selection bias (group allocation linked to reconstruction feasibility	

JBI Critical Appraisal Checklist for Case Series	
Citation: Zawawi, F., Bukhari, A. F., Khairy, S. A., & Garrada, M. (2025). Long-term clinical and audiometric outcomes after the implantation of piezoelectric bone conduction devices in children: a prospective cohort study. European Archives of Oto-Rhino-Laryngology. doi:10.1007/s00405-025-09339-3	
Checklist item	**Appraisal**
1. Were there clear criteria for inclusion in the case series?	Yes
2. Was the condition measured in a standard, reliable way for all participants included in the case series?	Yes
3. Were valid methods used for identification of the condition for all participants included in the case series?	Yes
4. Did the case series have consecutive inclusion of participants?	Yes
5. Did the case series have complete inclusion of participants?	Yes
6. Was there clear reporting of the demographics of the participants in the study?	Yes
7. Was there clear reporting of clinical information of the participants?	Yes

8. Were the outcomes or follow-up results of cases clearly reported?	Yes
9. Was there clear reporting of the presenting site(s)/clinic(s) demographic information?	Yes
10. Was statistical analysis appropriate?	Yes
Overall risk of bias	Low
• Strengths: Prospective design, consecutive inclusion, comprehensive outcome measures, long follow-up. • Limitations: Single centre, single surgeon; small sample size (n=25)	

JBI Critical Appraisal Checklist for Case Series	
Citation: Cushing, S. L., Gordon, K. A., Purcell, P. L., Feness, M., Negandhi, J., & Papsin, B. C. (2022). Surgical Considerations for an Osseointegrated Steady State Implant (OSIA2 R) in Children. Laryngoscope, 132(5), 1088-1092. doi:https://dx.doi.org/10.1002/lary.29892	
Checklist item	**Appraisal**
1. Were there clear criteria for inclusion in the case series?	Yes
2. Was the condition measured in a standard, reliable way for all participants included in the case series?	Yes
3. Were valid methods used for identification of the condition for all participants included in the case series?	Yes
4. Did the case series have consecutive inclusion of participants?	Not specified
5. Did the case series have complete inclusion of participants?	Yes
6. Was there clear reporting of the demographics of the participants in the study?	Yes
7. Was there clear reporting of clinical information of the participants?	No
8. Were the outcomes or follow-up results of cases clearly reported?	Yes
9. Was there clear reporting of the presenting site(s)/clinic(s) demographic information?	Yes
10. Was statistical analysis appropriate?	N/A
Overall risk of bias	Low to moderate
• **Strengths**: Clear description of surgical technique and complications. • **Limitations**: Outcomes largely descriptive (no audiological outcomes – surgical focus), method of follow-up not specified (unclear if complications are short or long-term, comorbidities not reported	

JBI Critical Appraisal Checklist for Cohort Studies	
Citation: Cushing, S. L., Goh, S., Treble, A., Papsin, B. C., & Gordon, K. A. (2024). Feasibility and Outcomes of an Active Osseointegrated Bone Conduction Implant in Children as Young as 5 Years of Age. Otology & Neurotology, 45(8), 913-918. doi:https://dx.doi.org/10.1097/MAO.0000000000004279	
Checklist item	**Appraisal**
1. Were the two groups similar and recruited from the same population?	Yes
2. Were the exposures measured similarly to assign people to both exposed and unexposed groups?	Yes
3. Was the exposure measured in a valid and reliable way?	Yes
4. Were confounding factors identified?	Yes
5. Were strategies to deal with confounding factors stated?	Limited – no multivariate analysis to adjust for comorbidities or aetiology
6. Were the groups/participants free of the outcome at the start of the study (or at the moment of exposure)?	Yes
7. Were the outcomes measured in a valid and reliable way?	Yes
8. Was the follow-up time reported and sufficient to be long enough for outcomes to occur?	Yes
9. Was follow-up complete, and if not, were the reasons to loss to follow-up described and explored?	Unclear – loss to follow-up not explicitly stated
10. Were strategies to address incomplete follow-up utilized?	Cannot fully exclude attrition bias
11. Was appropriate statistical analysis used?	Yes
Overall risk of bias	Low to moderate
• **Strengths**: large sample size, appropriate basic statistical comparisons • **Limitations**: lack of adjustment for potential confounders (aetiology, comorbidities), unclear reporting on f/u duration/completeness	

JBI Critical Appraisal Checklist for Case Series	
Citation: Gordon, K. A., Papsin, B. C., Feness, M., Negandhi, J., & Cushing, S. L. (2022). First Generation Osseointegrated Steady State Implant Benefits in Children With Hearing Loss. Otology & Neurotology, 43(3), 337-344. doi:10.1097/mao.0000000000003447	
Checklist item	**Appraisal**
1. Were there clear criteria for inclusion in the case series?	Yes
2. Was the condition measured in a standard, reliable way for all participants included in the case series?	Yes
3. Were valid methods used for identification of the condition for all participants included in the case series?	Yes
4. Did the case series have consecutive inclusion of participants?	Yes
5. Did the case series have complete inclusion of participants?	Yes
6. Was there clear reporting of the demographics of the participants in the study?	Yes
7. Was there clear reporting of clinical information of the participants?	Yes
8. Were the outcomes or follow-up results of cases clearly reported?	Yes
9. Was there clear reporting of the presenting site(s)/clinic(s) demographic information?	Yes
10. Was statistical analysis appropriate?	Yes
Overall risk of bias	Low
• **Strengths**: well-designed prospective case series, clearly defined eligibility criteria, systematic outcome collection, appropriate analysis. • **Limitations**: Industry-sponsored trial, single centre - may limit generalisability.	

JBI Critical Appraisal Checklist for Case Series	
Citation: Leonard, C., Oberoi, M., Belza, C., Carvalho, D., Gosman, A., & Lance, S. (2025). Single-Stage Ear Reconstruction Simultaneous With Piezoelectric Osia R Bone Conduction Implantation. Annals of Plastic Surgery, 94(5S Suppl 3), S441-S445. doi:https://dx.doi.org/10.1097/SAP.0000000000004280	
Checklist item	**Appraisal**
1. Were there clear criteria for inclusion in the case series?	Yes
2. Was the condition measured in a standard, reliable way for all participants included in the case series?	Yes
3. Were valid methods used for identification of the condition for all participants included in the case series?	Yes
4. Did the case series have consecutive inclusion of participants?	Unclear
5. Did the case series have complete inclusion of participants?	Yes
6. Was there clear reporting of the demographics of the participants in the study?	Yes
7. Was there clear reporting of clinical information of the participants?	Yes
8. Were the outcomes or follow-up results of cases clearly reported?	Yes
9. Was there clear reporting of the presenting site(s)/clinic(s) demographic information?	Yes
10. Was statistical analysis appropriate?	N/A
Overall risk of bias	Low
• Strengths: prospective design, explicit eligibility/exclusion, detailed surgical technique and outcomes, >2 years follow-up. • Limitations: unclear consecutive recruitment, small sample size, limited outcome reporting (audiological data not included in this publication), descriptive only.	

Appendix 3: Audiological Outcome Measurement Methods

Author	Conditions	PTA Calculation	Functional Gain Calculation	WRS or SRT Quiet	Speech-in-noise	Other / notes
Mozaffari 2025 (72)	Unaided	Pure tone thresholds 0.5-4 kHz	N/A	N/A	Average speech recognition scores (% correct) at 0 dB, 5 dB and 15 dB SNR	SSD excluded from audiometric analysis (n = 44/49 included) Summary figures provided only
You 2022 (66)	Unaided	PTA4 0.5-3 kHz	Improvement in AC PTA at 0.5-3 kHz	SRT in quiet WRS - Various age-based speech discrimination tests (NU-6, AzBio, WIPI, CNC, PB-K, HINT)	N/A	All were CMHL patients
Florentine 2022 (67)	Unaided, Aided (assorted devices)	PTA4 0.5-4 kHz	N/A	N/A	N/A	Subgroup analysis for aural atresia/SSD Results not paired
Stevens 2025 (68)	Unaided, Baha 5 Power on Softband	PTA4 0.5-4 kHz	N/A	CNC word scores at 60 dBA	BKB-SIN, SNR-50 (SNR threshold at which 50% of words correct at 65 dBA)	CMHL/SSD not separated
Zawawi 2025 (70)	Unaided	Median PTA 0.25-8 kHz	N/A	Monosyllabic speech discrimination test at 40 dB (not compared to pre-op)	N/A	Sound localisation testing Subgroup analysis for standard vs posterior placement CMHL patients only
Zawawi 2025 (69)	Unaided	Median PTA 0.25–8 kHz	N/A	Monosyllabic speech discrimination test	N/A	Subgroup analysis for CHL and SSD
Gordon (76)	Unaided	PTA4 0.5–4 kHz	N/A	% correct at 65 dB SPL using PB-K word list	N/A	CMHL/SSD not separated

AC = Air Conduction; AzBio = Arizona Biomedical Sentences; CHL = Conductive Hearing Loss; CMHL = Conductive or Mixed Hearing Loss; CNC = Consonant-Nucleus-Consonant words lists; HINT = Hearing in Noise Test; N/A = Not Applicable/Not Reported; NU-6 = Northwestern University Auditory Test No. 6; PB-K = Phonetically Balanced Kindergarten word lists; PTA = Pure Tone Average; SIN = Speech-in-noise; SNR = Signal to Noise Ratio; SRT = Speech Reception Threshold; SSD = Single-Sided Deafness; WIPI = Word Intelligibility by picture Identification.; WRS = Word Recognition Score

Appendix 4: Ethical Approval

Address for all correspondence
Research Ethics and Governance Office
Royal Prince Alfred Hospital

Telephone: (02) 9515 6766
Email: SLHD-RPAEthics@health.nsw.gov.au
Reference: X25-0154 & 2025/ETH00417

Health
NSW GOVERNMENT | Sydney Local Health District

01 August 2025

Dear A/Prof P Mukherjee,

Re: Protocol No. X25-0154 & 2025/ETH00417 - "Outcomes of Active Bone Conduction Hearing Devices in Children with Congenital Aural Atresia"

Thank you for submitting the above research proposal for single ethical and scientific review. This project was first considered by the Executive of the Sydney Local Health District Human Research Ethics Committee (HREC) – RPAH Zone at its meeting held on 29 July 2025.

This HREC is constituted and operates in accordance with the National Health and Medical Research Council's *National Statement on Ethical Conduct in Human Research*, the *CPMP/ICH Note for Guidance on Good Clinical Practice* and the *National Clinical Trials Governance Framework*.

I am pleased to advise that final ethical approval has been granted based on the following:

- The research project meets the requirements of the *National Statement on Ethical Conduct in Human Research (2023)*
- The HREC granted a waiver of the usual requirement for the consent of the individual for the use of their health information in a research project, in accordance with the Guidelines approved under Section 95A of the *Privacy Act 1988*. You are therefore required to keep detailed records of the following: the name(s) of the private sector organisation(s) from which health information was collected, the data fields collected and the number of records accessed for this research project. This information will be reported annually to the Commonwealth Privacy Commissioner via the Australian Health Ethics Committee.

The documents reviewed and approved include:

- HREA (Version 2, 21 July 2025)
- Protocol (Version 1.1, 21 July 2025)
- Data Variables List (Version 1.0, 26 May 2025)
- Master Code Sheet (Version 1, 26 February 2025)

- SLHD Privacy Compliance Form (Version 3, October 2020)
- Research Data Management Plan (Version 1, 28 July 2025)

The HREC has provided ethical and scientific approval for the following sites:

1. Royal Prince Alfred Hospital, NSW
2. Surgical rooms of A/Prof Payal Mukherjee:
   ENT Care Sydney
   Suite 210, SAN Clinic Tuloch
   185 Fox Valley Road 2076

For sites outside of NSW/ACT REGIS, a copy of this letter must be forwarded to all site investigators for submission to the relevant Research Governance Officer.

Please note the following conditions of approval. The conditions listed in this approval letter should be comprehensively reviewed and understood by all members of the research team:

1. HREC approval is valid for five (5) years subject to the supply of annual progress reports. The first report should be sent to the HREC by **01 August 2026.** You must also provide an annual report to the HREC upon completion of the study. This will be through a submission of a milestone in REGIS, see REGIS Quick Reference Guide (QRG): Submitting Annual Progress or Final Report (Milestone).

   Important notes:
   - **Ethics expiry:** An ethics extension amendment should be submitted prior to the ethics approval expiry date if the study is continuing beyond that date. This will be through a notification of an ethics amendment via REGIS, see REGIS QRG: Ethics Amendment - Completing and Submitting. Projects that are 12 months past the ethics expiry without submitting an ethics extension amendment will automatically be **suspended**.
   - **Milestones:** The status of any pending annual progress report that is six or more months past the due date will automatically be changed to 'Not Achieved'. The Research Office should be contacted to create a replacement milestone for the calendar year covered by the 'Not Achieved' milestone. The Committee relies on these reports to verify that the conduct of research complies with the approved protocol and remains ethically acceptable. Failure to submit regular or ongoing reports may result in your **ethics approval being withdrawn.**
2. In accordance with the National Statement, chapter 4.7; you must seek ethical approval from the HREC of the Aboriginal Health and Medical Research Council (AHMRC) if you intend to use Aboriginal and /or Torres Strait Islander status in any presentation or publication. See Research Office website for more information.
3. The study procedures as listed in the protocol must be followed at all times. See *The Australian Code for the Responsible Conduct of Research*.
4. All study personnel must be trained in the study protocol and aware of their role and responsibilities with respect to the research. All new personnel must be appropriately onboarded.
5. **Ethics Amendments:** Any proposed changes to the research protocol should be submitted to the HREC before those changes are implemented, such as changes to:
   - The general conduct of the research, including new aims or sub-studies

   - Any study procedures or data collection/management
   - CPI, site PI, adding students or other study personnel
   - The period covered by the ethics approval, i.e, to request an extension
   - The addition of sites

   Updated study documents should be submitted as a tracked and clean copy with new version number and date. This will be through a notification of an ethics amendment via REGIS, see REGIS QRG: Ethics Amendment - Completing and Submitting. See the Research Office website for more information on who can submit an amendment.
6. If the project is discontinued at a site before the expected date of completion, you must notify the HREC with reasons provided. It is also important to ensure study closure and completion processes are carried out in accordance with the Research Data Management Plan, Good Clinical Practice and local governance procedures. This will be through a notification of an ethics amendment via REGIS, see REGIS QRG: Ethics Amendment - Completing and Submitting. The site Research Governance Officer should also be notified following ethics acknowledgment, see REGIS QRG: Governance Amendment - Completing and Submitting.
7. You must immediately report anything which might warrant review of ethics approval, including unforeseen events that might affect continued ethical acceptability of the project. Examples include, significant safety issues, serious breaches, participant complaints, privacy breaches. This will be through a notification via REGIS, see REGIS QRG: Clinical Trial Safety Reporting (for clinical trials) or Ethics Amendment - Completing and Submitting.
8. **Serious breaches:** Serious breaches and complaints should be reported in accordance with NHMRC Guidance document: Reporting of Serious Breaches of Good Clinical Practice (GCP) or the Protocol for Trials Involving Therapeutic Goods 2018. **All complaints should immediately be reported to the HREC within 24 hours of being notified.** This will be through a notification via REGIS. This will be through a notification via REGIS, see REGIS QRG: Clinical Trial Safety Reporting (for clinical trials) or Ethics Amendment - Completing and Submitting.
9. **Conflicts of interest:** Any changes to financial, business or other non-financial conflicts of interests related to this research should be declared to the HREC in accordance with the National Statement Chapter 5.4: Conflicts of Interest. See also NHMRC guidance document Disclosure of interests and management of conflicts of interest. This will be through a notification via REGIS, see REGIS QRG: Ethics Amendment - Completing and Submitting.
10. **Student projects:** HREC approval is granted on the assumption that all students and early career researchers are adequately supervised by the Coordinating Principal Investigator and senior investigators on a project. This supervision would ensure that all privacy concerns are met (including the completion of confidentiality agreements by participating students) and that students are supported in the conduct of the study in line with the approved research protocol.

For your information at the end of this letter is a general checklist to assist you with following all the necessary steps to support the study's compliance throughout its full duration.

Should you have any queries about the HREC's consideration of your project please contact the Executive Officer - (02) 9515 6766. The HREC Terms of Reference, Standard Operating Procedures, membership and standard forms are available from the website: https://www.slhd.nsw.gov.au/rpa/research/default.html

The HREC welcomes feedback from researchers on how the ethics review process can be improved or how researchers can be better supported. If you would like to provide feedback, please email the Research Office.

Researchers are encouraged to:
- Develop standard operating procedures for consenting in line with the National Standard Operating Procedures. (if applicable)
- Regularly visit REGIS for system updates and for notifications about their project.
- Regularly review the Research Office website for up-to-date information on ethics requirements, training opportunities and drop-in clinics.
  https://www.slhd.nsw.gov.au/rpa/research/default.html

The Human Research Ethics Committee wishes you every success in your research.

Yours sincerely,

Executive Officer
Sydney Local Health District Human Research Ethics Committee – RPAH Zone