

Special Issue Reprint

Diagnosis and Management of Pediatric Ear and Vestibular Disorders

Edited by
Mirko Aldè and Stefania Barozzi

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Diagnosis and Management of Pediatric Ear and Vestibular Disorders

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Guest Editors

Mirko Aldè

Stefania Barozzi



Basel • Beijing • Wuhan • Barcelona • Belgrade • Novi Sad • Cluj • Manchester

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About the Editors

Mirko Aldè

Mirko Aldè is a medical doctor specializing in Audiology and Phoniatrics. He currently practices at the Fondazione IRCCS Ca' Granda Ospedale Maggiore Policlinico in Milan and serves as a researcher at the University of Milan, Italy. His clinical and research activities focus on the diagnosis and management of hearing and balance disorders, with particular attention to pediatric populations. His primary interests include pediatric hearing loss and vestibular dysfunction associated with otitis media, genetic conditions, congenital cytomegalovirus infection, and prematurity. He is actively involved in advancing early detection and intervention strategies aimed at improving long-term auditory and developmental outcomes in children.

Stefania Barozzi

Stefania Barozzi is a medical doctor specializing in Audiology and an Associate Professor at the University of Milan, Italy. She has extensive clinical and research experience in audiological and vestibular disorders, with particular expertise in the diagnosis and management of Ménière's disease, persistent postural-perceptual dizziness (PPPD), benign paroxysmal positional vertigo (BPPV), vestibular neuritis, tinnitus, and hearing loss. Her research focuses on improving diagnostic approaches and treatment strategies for patients with hearing and balance disorders, with the aim of enhancing clinical outcomes and quality of life.

Preface

Disorders affecting hearing and balance during childhood present unique clinical challenges because of their potential impact on communication, motor development, learning, and quality of life. Continued progress in diagnostic methods and therapeutic approaches has expanded the opportunities for earlier recognition and more effective management of these conditions.

This Reprint reflects the breadth of contemporary research in pediatric audiology and vestibular medicine through a collection of original studies and reviews covering a wide range of clinical topics. The contributions offer practical insights, discuss unresolved issues, and highlight areas where further investigation is needed.

We hope that this Reprint will provide a valuable resource for clinicians, researchers, and healthcare professionals with an interest in pediatric hearing and balance disorders, while fostering further advances in both research and clinical practice.

Mirko Aldè and Stefania Barozzi

Guest Editors

Diagnosis and Management of Pediatric Ear and Vestibular Disorders

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Hearing and balance disorders represent some of the most challenging conditions encountered in pediatric healthcare [1]. Although substantial progress has been achieved in newborn hearing screening, audiological assessment, and otologic surgery over recent decades, many children with auditory and vestibular dysfunction continue to experience delayed diagnosis, limited access to specialized care, and significant impacts on communication, learning, motor development, and quality of life [2,3]. The complexity of these disorders is amplified by the unique developmental characteristics of childhood, which often make symptom recognition and clinical evaluation more difficult than in adult populations [4].

The Special Issue “*Diagnosis and Management of Pediatric Ear and Vestibular Disorders*” was designed to highlight current challenges and emerging opportunities in this important yet still understudied field, with contributions addressing the diagnosis of vestibular disorders, the diagnosis and treatment of hearing loss (HL), innovative rehabilitation approaches such as telerehabilitation and music-based interventions, and the management of complications related to otitis media. Overall, nine original articles and one review are included in this Special Issue, as listed below:

1. Božanić Urbančič, N.; Mladenov, D.; Battelino, S. The Impact of Follow-Up on Etiological Classification of Pediatric Vertigo. *Children* **2026**, *13*, 376. <https://doi.org/10.3390/children13030376>
2. Norton, C.P.; Darr, N.S.; Beshears, M.K.; Catalano, K.; Dillard, T.; Gamble, M.J.K.; Olerich, M.; Rupp, S.R. Non-Instrumented DVA: Assessment of Performance and Clinical Feasibility in Children Ages 2 Through 13 Years. *Children* **2026**, *13*, 456. <https://doi.org/10.3390/children13040456>

Specifically, it was shown that longitudinal vestibular follow-up should be regarded as an essential component of the diagnostic pathway, facilitating greater diagnostic accuracy and a more comprehensive understanding of the underlying disorder. Alongside this, the feasibility of non-instrumented dynamic visual acuity (DVA) testing in children was demonstrated, supporting the use of a simple and accessible screening tool that may facilitate the early identification of vestibular dysfunction in routine clinical practice.

The studies on auditory disorders mainly focused on various aspects of diagnosis, rehabilitation, and treatment, and the contributions are listed below:

1. Brotto, D.; Impalà, G.; Lovato, E.; Mazzaro, E.; Maculan, M.; Zanoletti, E.; Galoforo, N.; Trevisi, P. ABR Features in Ski-Slope Hearing Loss for Hearing Threshold Estimation: A Comparative Clinical Study of Click and CE-Chirp Stimuli. *Children* **2026**, *13*, 410. <https://doi.org/10.3390/children13030410>

2. Aldè, M.; Zanetti, D.; Ambrosetti, U.; Monaco, E.; Gasbarre, A.M.; Pignataro, L.; Cantarella, G.; Barozzi, S. Unilateral Sensorineural Hearing Loss in Children: Etiology, Audiological Characteristics, and Treatment. *Children* **2024**, *11*, 324. <https://doi.org/10.3390/children11030324>.
3. Lauriello, M.; Angelone, A.M.; Iannotti, S.; Nardecchia, E.; Scopano, B.; Fioretti, A.; Ciancarelli, I.; Eibenstein, A. Audiophonologopedic Telerehabilitation: Advantages and Disadvantages from User Perspectives. *Children* **2024**, *11*, 1073. <https://doi.org/10.3390/children11091073>.
4. Aldè, M.; Casella, L.; Ambrosetti, U.; Barozzi, S.; Gandolfo, E.; Di Berardino, F.; Zanetti, D. Music-Based Interventions in Childhood Hearing Loss: A Comprehensive Narrative Review. *Children* **2026**, *13*, 574. <https://doi.org/10.3390/children13040574>.

It was reported that, in the context of auditory brainstem responses (ABRs), CE-Chirp stimulation offers clearer advantages in threshold estimation and neural synchrony, particularly in pediatric patients with ski-sloping HL. Moreover, children with unilateral HL were shown to perform significantly better on audiological tests, including speech perception in quiet and noisy environments and sound localization, while using hearing devices. These findings suggest that greater attention should be devoted to the timely identification and rehabilitation of these patients. Finally, telerehabilitation and music-based interventions emerged as valuable tools for children with HL, representing feasible approaches for future rehabilitation strategies.

Another important topic addressed in this Special Issue was the management of complications of otitis media, with contributions exploring endoscopic myringoplasty, the pediatric utilization of ophthalmic antibiotics in the ear, and the clinical management of acute mastoiditis, as listed below:

1. Nocini, R.; Monzani, D.; Arietti, V.; Bonasera, F.; Bianconi, L.; Sacchetto, L. Endoscopic Myringoplasty for Pediatric Tympanic Membrane Perforations: Is It Worth It? *Children* **2025**, *12*, 293. <https://doi.org/10.3390/children12030293>.
2. Cheung, I.M.Y.; Yin, T.; Gokul, A. Paediatric Utilisation of Ophthalmic Antibiotics in the Ear in Aotearoa/New Zealand. *Children* **2025**, *12*, 557. <https://doi.org/10.3390/children12050557>.
3. Alshehri, S.; Alahmari, K.A. Pediatric Acute Mastoiditis in Saudi Arabia: Demographic Insights, Clinical Profiles, and Prognostic Factors. *Children* **2024**, *11*, 402. <https://doi.org/10.3390/children11040402>.
4. Sabitova, N.; Shamshudinov, T.; Kussainova, A.; Toguzbayeva, D.; Sadykov, B.; Rahanskaya, Y.; Kassym, L. A National Forecast and Clinical Analysis of Pediatric Acute Mastoiditis in Kazakhstan. *Children* **2026**, *13*, 170. <https://doi.org/10.3390/children13020170>.

Endoscopic myringoplasty was shown to be a safe procedure with a high success rate for the repair of tympanic membrane perforations, while further collaborative efforts are needed to optimize the appropriate use of ophthalmic ciprofloxacin for ear infections. The studies from Saudi Arabia and Kazakhstan demonstrated that pediatric acute mastoiditis is both a clinically significant complication of otitis media and a sensitive indicator of broader healthcare system constraints, underscoring the need for earlier diagnosis, standardized management pathways, and strengthened pediatric otolaryngology services to prevent severe complications and improve outcomes.

Overall, this Special Issue provides novel insights and has the potential to contribute significantly to the diagnosis, management, treatment, and rehabilitation of hearing and vestibular disorders in children. Future research should focus on the implementation of universal vestibular screening programs for children with HL [5], the development of more reliable diagnostic tools for vestibular disorders in early childhood [3,4], and the

advancement of innovative hearing and vestibular rehabilitation strategies [6]. Particular attention should be devoted to ensuring that technologically advanced diagnostic and rehabilitative tools become accessible to pediatric populations worldwide [7].

Author Contributions: Conceptualization, M.A.; methodology, M.A.; writing—original draft preparation, M.A.; writing—review and editing, M.A. and S.B.; visualization, S.B.; supervision, S.B. All authors have read and agreed to the published version of the manuscript.

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Conflicts of Interest: The authors declare no conflicts of interest.

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3. Aldè, M.; Fancello, V.; Di Mauro, P.; Canelli, R.; Zaouche, S.; Falanga, C. Audiological and Vestibular Follow-Up for Children with Congenital Cytomegalovirus Infection: From Current Limitations to Future Directions. *Children* **2024**, *11*, 1211. [CrossRef] [PubMed]
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Article

The Impact of Follow-Up on Etiological Classification of Pediatric Vertigo

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Highlights

What are the main findings?

- Extended longitudinal follow-up dramatically reduced the proportion of pediatric vertigo with initially unknown etiology, from 43% at first evaluation to 10% after diagnostic reassessment.
- Many cases initially classified as unexplained vertigo evolved into identifiable clinical entities, most commonly migraine-related vertigo, as well as central, hemodynamic, psychogenic, and peripheral vestibular disorders.

What are the implications of the main findings?

- In pediatric vertigo, follow-up should be regarded as an active diagnostic tool, not merely observation, because clinical phenotypes often emerge over time.
- Early diagnostic uncertainty is common in children with vertigo and should prompt structured longitudinal evaluation rather than premature diagnostic closure.

Abstract

Background: Vertigo and dizziness in children represent diagnostically challenging conditions with heterogeneous etiologies. At initial presentation, a substantial proportion of pediatric patients remain without a definitive etiological diagnosis. Evidence on the impact of longitudinal follow-up on etiological classification in pediatric vertigo is limited. **Methods:** This observational cohort study uses prospectively collected clinical data. Children aged 1–17 years who presented to a tertiary ENT clinic with vertigo and/or dizziness between 2015 and 2020 were systematically enrolled and followed. The present study represents a retrospective revision of a previously published cohort of 257 children. In 2025, extended follow-up data were reviewed to reassess etiological classification using the same diagnostic categories as in the original analysis. Descriptive statistics were applied to compare etiological distributions at initial evaluation and after follow-up revision. **Results:** After data revision, the proportion of children with unclassified etiology decreased from 44% to 10%. Central etiologies accounted for 35% of cases, peripheral vestibular disorders for 18%, hemodynamic causes for 16%, psychogenic etiologies for 10%, and other specific causes for 7%. Follow-up duration ranged from 0 to 132 months (mean 17.6 months; median 4.5 months). Diagnostic investigations were frequently performed; however, the etiological yield of certain tests, particularly cranial computed tomography, was low. **Conclusions:** Extended follow-up significantly improves etiological classification in children with vertigo and dizziness, demonstrating that diagnostic uncertainty at initial presentation often reflects evolving clinical phenotypes rather than the absence of an underlying disorder. A longitudinal, clinically guided, and multidisciplinary approach is

essential to enhance diagnostic accuracy and optimize the use of diagnostic investigations in pediatric vertigo.

Keywords: pediatric vertigo; dizziness; follow-up; etiology; vestibular disorders; migraine

1. Introduction

Vertigo and dizziness in childhood represent complex clinical entities with diverse etiologies, spanning central vestibular disorders, peripheral vestibulopathies, systemic factors, and psychogenic components. Although pediatric vertigo is less commonly encountered than in adults, its impact on quality of life, cognitive function, and psychosocial development can be substantial if not appropriately diagnosed and managed [1,2].

Epidemiological studies indicate that migraine-related syndromes—including vestibular migraine and its pediatric variants such as benign paroxysmal vertigo of childhood (BPVC), now often termed recurrent vertigo of childhood—are among the most frequent causes of vertigo in children, followed by peripheral vestibular disorders and less common conditions such as vestibular neuritis and psychogenic vertigo [1,3,4].

Benign paroxysmal vertigo of childhood, a condition frequently linked to migraine spectrum disorders, is characterized by episodic vertiginous spells in the absence of a persistent vestibular lesion and is among the most common identifiable causes of vertigo in young children [5,6]. The International Classification of Headache Disorders recognizes these migraine-related variants, reflecting their central neurological origin and often overlapping clinical features with migraine itself [2].

Despite advances in diagnostic techniques and increasing awareness among clinicians, a substantial proportion of pediatric patients still present with vertigo or dizziness that cannot be definitively classified at initial presentation. Large retrospective series have reported that approximately 20% of cases have an undetermined etiology after the first clinical evaluation [4]. This diagnostic uncertainty poses challenges for prognostication, therapeutic decision-making, and patient counseling.

Longitudinal follow-up studies in pediatric vestibular populations are comparatively scarce, yet evidence suggests that etiological classifications may evolve over time, particularly as clinical features mature, new symptoms emerge, or additional diagnostic information becomes available [7]. Such temporal evolution is especially notable for conditions like recurrent vertigo of childhood and vestibular migraine, where initial presentations may not fully meet diagnostic criteria but subsequently develop into more defined syndromes.

In our previously reported cohort from a tertiary referral center, a significant proportion of children presenting with vertigo and dizziness were classified as having an unexplained etiology following multidisciplinary assessment [8]. In most cases, follow-up evaluation was deemed clinically necessary and therefore scheduled at the initial visit. After systematic revision of the cohort in 2025, incorporating extended clinical follow-up and updated diagnostic information, we observed that many children initially labeled as unclassified had subsequently received specific diagnoses. These findings underscore the dynamic nature of pediatric vertigo and highlight the diagnostic value of longitudinal evaluation.

Therefore, the present study aims to re-evaluate the etiological distribution in our pediatric vertigo cohort after extended follow-up, focusing on changes in classification and the implications for clinical practice. By comparing initial and follow-up etiological assignments, we seek to quantify how longitudinal data influence diagnostic accuracy and to inform recommendations for follow-up strategies in pediatric vestibular disorders.

2. Materials and Methods

2.1. Study Design and Population

This study is an observational cohort study based on prospectively collected clinical data. Children presenting with vertigo and dizziness have been systematically enrolled and followed since 2015. The present analysis represents a retrospective review and revision of etiological classification using extended follow-up data.

The original cohort included consecutive children and adolescents aged 1–17 years who presented with vertigo and/or dizziness and were referred for multidisciplinary evaluation between 2015 and 2020.

In 2025, a systematic collection and revision of the clinical data was conducted, incorporating additional follow-up information collected after the initial publication. The aim of the revision was to reassess etiological classification in light of newly available diagnostic information that emerged during long-term follow-up.

No new patients were added to the cohort. The study population, therefore, corresponds exactly to the cohort described in the original publication.

2.2. Diagnostic Workup and Follow-Up

All children underwent a comprehensive diagnostic evaluation at initial presentation, as described previously, including detailed medical history, otoneurological examination, age-appropriate audiological and vestibular testing, and referral to relevant pediatric subspecialists (pediatric neurology, cardiology, psychology/psychiatry, ophthalmology, infectious disease, and radiology) based on the clinical presentation. Follow-up was scheduled in children with: persistent or recurrent symptoms, undefined etiology, suspected migraine phenotype, or abnormal neurological or vestibular findings. No strictly standardized intervals were applied; follow-up was clinically tailored.

Follow-up data of regular check-ups were obtained from medical records documenting subsequent outpatient visits, hospital admissions, specialist consultations, and diagnostic investigations performed after the initial evaluation. Particular attention was paid to children who were originally classified as having vertigo and dizziness of unclassified etiology.

Vestibular assessment was adapted to the child's age and level of cooperation and included bedside examination and, when feasible, instrumental testing such as video head impulse testing (vHIT-Interacustics, Middelfart, Denmark), caloric testing (Variotherm plus ATMOS MedizinTechnik GmbH and Co. KG Ludwig-Kegel, Lenzkirch, Germany), and vestibular evoked myogenic potentials (cVEMP; Eclipse, Interacustics A/S, 6610 Assens, Denmark). In younger children, completion of instrumental testing was occasionally limited by cooperation and developmental factors.

During follow-up, etiological reclassification was performed when sufficient clinical, diagnostic, or specialist-confirmed information became available to support a specific diagnosis. Diagnoses established during follow-up were based on accepted clinical criteria relevant at the time of reassessment (e.g., migraine-related disorders, autonomic/hemodynamic disorders, central neurological conditions, psychological disorders). Migraine-related vertigo was diagnosed according to Bárány Society and ICHD-3 criteria. BPPV was diagnosed based on positional nystagmus consistent with canal involvement. Vestibular neuronitis was defined by acute, prolonged vertigo, compatible vestibular findings, and the absence of central signs.

2.3. Etiological Classification

Etiological categories were identical to those used in the original study to allow direct comparison between initial and revised classifications. These included: central causes,

peripheral vestibular causes, hemodynamic causes, psychological causes, other identified causes, and unclassified etiology.

Reclassification was conducted by reviewing follow-up documentation and specialist reports. A diagnosis was considered revised only when supported by documented clinical assessment and/or diagnostic testing performed during follow-up. Psychogenic etiologies were diagnosed after exclusion of organic causes and, when indicated, in collaboration with pediatric neurology or psychology services. No diagnosis was made based solely on the absence of findings.

Serological testing for *Borrelia burgdorferi* was performed only when clinically indicated (e.g., compatible neurological symptoms, tick exposure, or endemic area).

2.4. Data Analysis

Descriptive statistics were used to summarize demographic characteristics and etiological distributions at initial evaluation and after follow-up revision. Categorical variables were expressed as frequencies and percentages. Continuous variables were summarized using means and standard deviations or medians and ranges, as appropriate.

Comparisons between initial and revised etiological classifications were performed to quantify changes in diagnostic distribution, with particular focus on the proportion of children initially classified as having an unclassified etiology who subsequently received a definitive diagnosis.

Statistical analysis was done using SPSS V20.0 (IBM, Armonk, NY, USA) and Microsoft Excel 2019 (Microsoft, Redmond, WA, USA). $p < 0.05$ was considered statistically significant.

3. Results

3.1. Study Population

A total of 257 children were included in the revised analysis. The cohort consisted of 42% males and 58% females, identical to the previously published population. The age range and demographic characteristics remained unchanged, as no new patients were added.

3.2. Overall Etiological Distribution After Revision (2025)

After extended clinical follow-up and evaluation of collected data, the etiology of vertigo and dizziness was reclassified in the majority of children. Most reclassifications occurred within the first two follow-up visits.

In total, 26 children (10%) remained classified as having unclassified etiology (Figure 1).

Figures 2 and 3 illustrate the distribution of peripheral and central etiologies after data revision. Among peripheral causes, benign paroxysmal positional vertigo predominated, while migraine-related vertigo represented the most frequent central diagnosis.

3.3. The Follow-Up Assessment

Follow-up data were available for 256 children. The duration of follow-up ranged from 0 to 132 months, with a mean follow-up time of 17.6 months, a median of 4.5 months, and an interquartile range (IQR) of 0–24 months. A follow-up duration of “0 months” indicates cases evaluated at a single clinical encounter with no subsequent scheduled reassessment.

Of the total cohort ($N = 257$), 52 children (20%) were not scheduled for follow-up, while 29 children (11%) did not attend the planned follow-up visit, resulting in 32% of children without a completed follow-up examination. Table 1 shows the extended multidisciplinary assessment during the follow-up. Neurological examination was performed in 190 children (74% of the total cohort). Of these, 164 children (86%) had normal neurological

status, while 26 (14%) had neurological abnormalities. EEG examination was performed in 94 children (36%). Normal EEG findings were observed in 85 children (90%), while 9 children (10%) showed pathological EEG findings. Tilt-table testing was performed in 13 children (5% of the total cohort). Normal findings were observed in 5 children (39%), while 8 children (62%) showed pathological results. The majority of cases were consistent with orthostatic intolerance and reflex-mediated mechanisms; formal autonomic testing was not systematically performed in all patients.

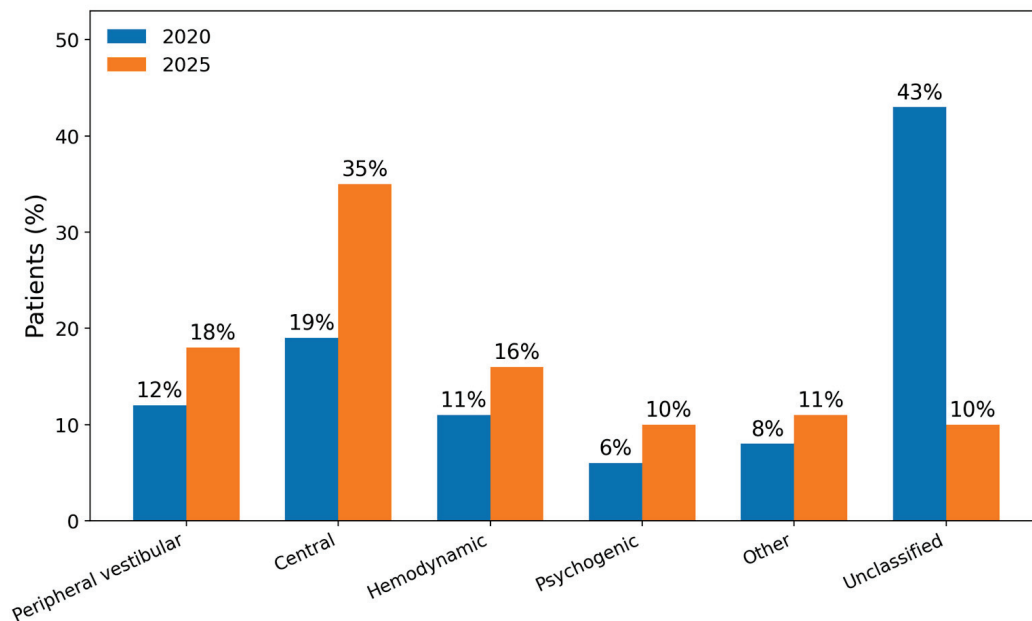


Figure 1. Comparison of etiological groups at initial evaluation (2020) and after follow-up revision (2025). Extended follow-up substantially reduced the proportion of unclassified cases and increased the identification of central, hemodynamic, psychogenic, and peripheral vestibular etiologies.

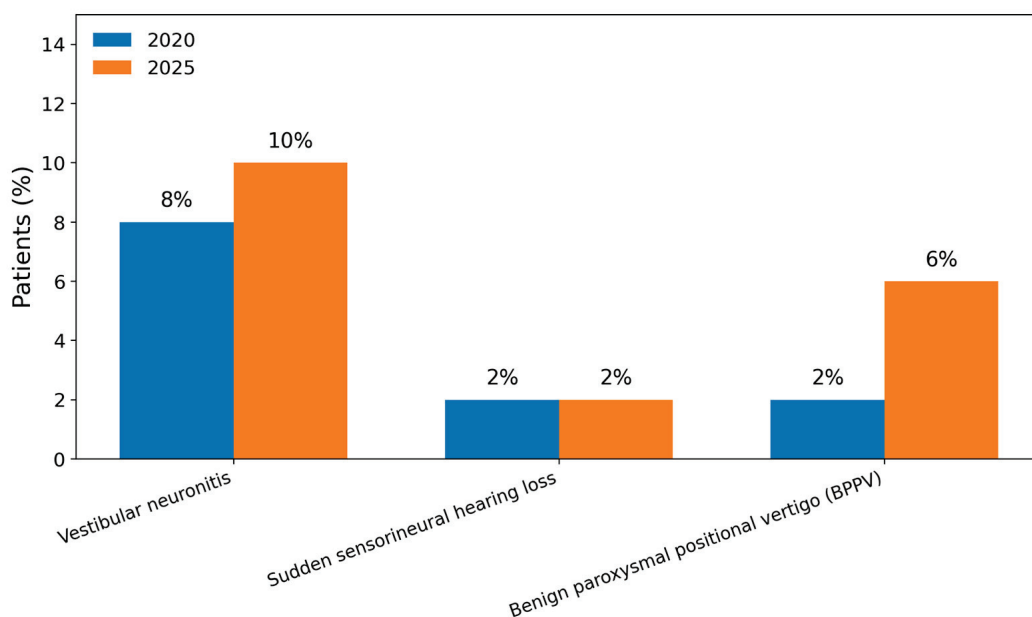


Figure 2. Etiological distribution of peripheral causes of pediatric vertigo and dizziness before (2020) and after data revision in 2025.

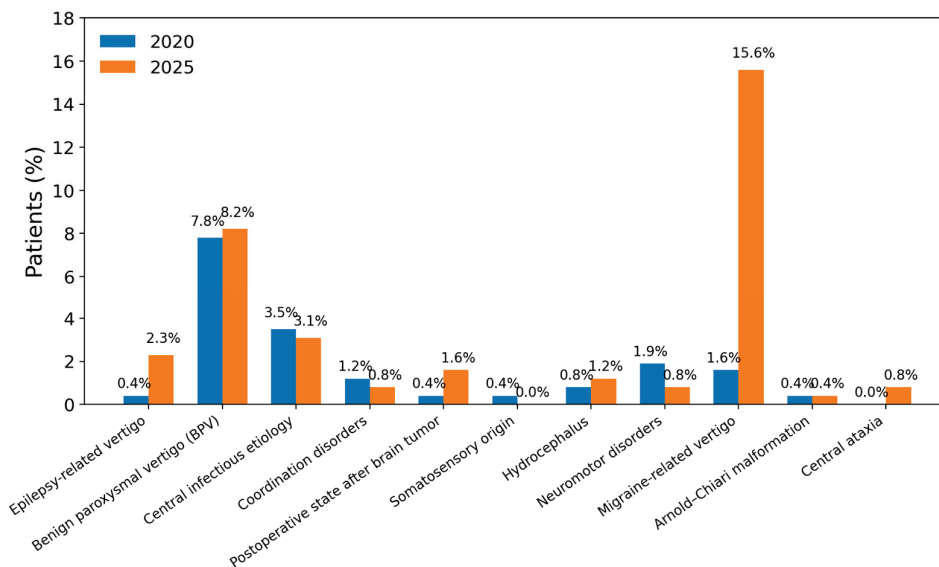


Figure 3. Distribution of central etiologies by diagnosis at initial evaluation (2020) and after follow-up revision (2025). Migraine-related vertigo emerged as the predominant central cause following extended follow-up, highlighting the diagnostic impact of longitudinal assessment.

Table 1. Diagnostic investigations and findings in the study cohort during the extended follow-up period. Percentages of normal and abnormal findings are calculated based on the number of children who underwent each specific diagnostic investigation.

Diagnostic Investigation	Children Examined, n (%)	Normal Findings, n (%)	Abnormal Findings, n (%)
Neurological examination	190 (74)	164 (86)	26 (14)
Electroencephalography (EEG)	94 (37)	85 (90)	9 (10)
Tilt-table test	13 (5)	5 (39)	8 (62)
Cranial computed tomography (CT)	160 (62)	158 (99)	2 (1)
Brain magnetic resonance imaging (MRI)	146 (57)	121 (83)	25 (17)
<i>Borrelia burgdorferi</i> serology	78 (30)	71 (91)	7 (9)
Ophthalmological examination	84 (33)	71 (85)	13 (15)

Cranial CT imaging was performed in 160 children (62% of the total cohort). Normal CT findings were observed in 158 children (99%), while 2 children (1%) showed pathological findings.

Brain MRI was performed in 146 children (57% of the total cohort). Normal MRI findings were observed in 121 children (83%), while 25 (17%) showed abnormal findings.

Serological testing for *Borrelia burgdorferi* was performed in 78 children (30% of the total cohort). Negative results were observed in 71 children (91%), while 7 children (9%) tested positive.

Ophthalmological examination or follow-up was performed in 84 children (33% of the total cohort). Of these, 13 children (15%) showed pathological ophthalmological findings.

4. Discussion

4.1. Longitudinal Reclassification of Etiology: Comparison Between 2021 and 2025

In contrast to our 2021 report, in which the etiology of vertigo and dizziness remained unclassified in 44% (112/257) of children [8], the 2025 data revision demonstrated a marked reduction in unclassified cases to 10% (26/257). This finding underscores that pediatric vertigo is frequently an evolving clinical entity, with diagnostic clarity often emerging only with longitudinal follow-up. Similar trends have been reported in recent pediatric cohorts, where extended observation and standardized diagnostic frameworks substantially reduced the proportion of unexplained cases [9,10]. Table 2. Demonstrates the percentage of unexplained pediatric vertigo and dizziness cases in different studies.

Table 2. Selected pediatric vertigo/dizziness studies reporting an unclassified/unknown etiology.

References	Year	Study Design/Setting	Sample (n)	Diagnostic Workup (Summary)	Unclassified/Unknown Etiology
Božanić Urbančič NB et al. Unraveling the Etiology of Pediatric Vertigo and Dizziness: A Tertiary Pediatric Center Experience. <i>Medicina (Kaunas)</i> . PMID: 34064850 [8]	2021	Retrospective cohort; tertiary pediatric ENT center	257	Multidisciplinary evaluation; clinical exam ± audiovestibular testing and targeted ancillary investigations	43.6% (112/257) unclassified
Wiener-Vacher SR et al. Epidemiology of Vestibular Impairments in a Pediatric Population. <i>Semin Hear</i> . PMID: 30038452 [4]	2018	Retrospective clinical series; referral center for complete vestibular testing	1037 (balance disorders subgroup)	Complete vestibular test battery (canal + otolith function) adapted for pediatrics	Unknown etiology 19.6% (among etiologies of balance disorders with vestibular impairment)
Khater A et al. Unveiling the Diagnosis of Pediatric Dizziness in a Tertiary Care Setting. PMID: 40291364 [11]	2025	Retrospective cohort; tertiary care	40	Clinical evaluation with vestibular/neurological assessment and ancillary tests as indicated (e.g., EEG, MRI)	22.5% (9/40) diagnosis not ascertained
Fancello V et al. Vertigo and Dizziness in Children: An Update. <i>Children (Basel)</i> . PMID: 34828738 [9]	2021	Systematic review (PRISMA) of recent pediatric vertigo studies	2470 (total across included studies)	Included heterogeneous diagnostic protocols across studies; summarizes etiologic distributions	Etiology unknown in 0.9% overall (as reported in review synthesis)
Davitt M; Delvecchio MT; Aronoff SC. The Differential Diagnosis of Vertigo in Children: A Systematic Review of 2726 Cases. <i>Pediatr Emerg Care</i> . 2020;36:368–371. doi:10.1097/PEC.0000000000001281 [1]	2020	Systematic review (2726 pediatric cases pooled)	2726	Across included studies, heterogeneous assessments (history/physical, bedside vestibular exam; variable audiology/imaging/lab/vestibular testing depending on source study)	Unidentified/idiopathic: 11.7% (credible interval reported in review).
Lorente-Piera J, et al. Clinical Profile, Trends, and Management in Pediatric Patients with Audiovestibular Disorders. <i>Audiol Res</i> . 2024. PMID: 39194415 [12]	2024	Retrospective cohort; pediatric ENT / audiovestibular setting	117	Audiovestibular evaluation; vHIT/VEMP in subset; imaging performed in 64.1% (diagnostic in 34.7%)	Idiopathic (no identified cause): 7.69% (n = 9).
Rey-Berenguel M, et al. Diagnostic Yield of the New Bárány Society Criteria for Pediatric Episodic Vestibular Syndrome. <i>J Clin Med</i> . 2025 [13]	2025	Observational cross-sectional; tertiary pediatric vertigo clinic; EVS focus	109 (EVS cohort)	Standardized neuro-otological exam; audiovestibular tests and imaging as needed; classification using ICHD-3 vs. Bárány 2021 criteria	Undetermined EVS without hearing loss reduced from 35.78% to 16.51% after applying Bárány 2021 criteria.

Recent single-center cohort studies have described the etiological spectrum of pediatric vertigo based predominantly on findings at initial presentation, without longitudinal reclassification; while these studies provide valuable cross-sectional insights, they do not capture the dynamic evolution of diagnoses over time, which may contribute to persistently high rates of unclassified cases [14]. In contrast, our longitudinal revision demonstrates that extended follow-up substantially improves etiological classification, supporting follow-up as an active diagnostic tool rather than a passive observational period.

While unclassified cases decreased, diagnoses within central, hemodynamic, psychogenic, and peripheral categories increased. Specifically, after the 2025 revision, central etiologies accounted for 35% (91/257) of cases, peripheral vestibular disorders for 18% (45/257), hemodynamic causes for 16% (41/257), psychogenic etiologies for 10% (26/257), and other specific etiologies for 7% (19/257), with only 10% remaining unclassified. On the other hand, considerable variability in epidemiological data across published studies likely reflects differences in study design, referral patterns, the professional backgrounds of investigators (ENT specialists, neurologists, pediatricians), diagnostic protocols, and the timing of evaluation. Furthermore, heterogeneity in bedside versus instrumental investigations and emergency versus outpatient assessment may substantially influence reported etiological distributions. These methodological differences partly explain discrepancies in the proportion of unclassified cases reported in the literature.

In comparison, the 2021 analysis reported substantially lower proportions for several of these categories, suggesting that initial diagnostic uncertainty often reflects incomplete phenotypic expression rather than the absence of an underlying disorder. This longitudinal shift mirrors findings from recent studies indicating that standardized classification and follow-up significantly improve etiological resolution in pediatric dizziness [15]. These findings further support the concept that pediatric vertigo represents a distinct clinical entity, requiring age-specific diagnostic pathways and longitudinal follow-up strategies, rather than extrapolation from adult data. Moreover, our findings support the concept of a “diagnostic waiting period” in pediatric vestibular medicine, in which longitudinal follow-up serves as an active diagnostic tool rather than passive observation. Symptom evolution over time allows more accurate etiological classification, particularly in migraine-related and developmentally evolving conditions.

4.2. The Follow-Up

The duration and structure of follow-up in our cohort reflect a clinically tailored approach rather than uniform longitudinal surveillance. Children who were not scheduled for follow-up represented cases in which comprehensive vestibular assessment could be completed at the initial visit and peripheral vestibular causes could be convincingly excluded based on clinical history, examination, and available testing. In contrast, children who were scheduled but did not attend follow-up visits may represent a heterogeneous group; in our healthcare setting, this is often interpreted as symptom resolution rather than disengagement from care, although objective confirmation of this assumption is not available. The absence of follow-up data in 32% of children highlights a system-level challenge in pediatric vestibular care. Structured recall protocols, predefined reassessment intervals for diagnoses that remain undefined, and improved parental counseling about the potential evolution of symptoms may improve longitudinal diagnostic accuracy. Importantly, extended follow-up proved critical for etiological clarification in a substantial proportion of patients, as diagnostic reclassification frequently occurred over time. This observation is consistent with previous reports, underscoring that delayed recognition of vestibular pathology is common in pediatric populations and supports the value of longitudinal reassessment [16].

4.3. Peripheral Etiologies

The increase in peripheral vestibular diagnoses observed after follow-up revision can be partly explained by the inherent challenges of pediatric vestibular assessment at the initial visit. Children do not always cooperate optimally, and comprehensive vestibular testing cannot consistently be completed during a single consultation, particularly in younger patients [17,18]. In addition, certain peripheral vestibular disorders such as benign paroxysmal positional vertigo (BPPV) may not be diagnosed at initial clinical presentation in pediatric patients, even when vestibular evaluation is performed, with diagnoses frequently established only after several weeks of symptom persistence and comprehensive follow-up evaluation, as shown in Brodsky's work on pediatric concussion cases in which peripheral disorders were recognized on average 133 days after symptom onset rather than at first assessment [16]. On the other hand, pediatric BPPV may resolve rapidly, particularly in younger children, which can result in underdiagnosis if vestibular assessment is not performed promptly. A structured and timely vestibular evaluation pathway is therefore essential to ensure accurate identification and reliable epidemiological data.

4.4. Central Etiologies

In contrast to the baseline analysis, migraine-related vertigo emerged as the most frequent central diagnosis after revision. This evolution is clinically plausible, as migraine-associated vestibular symptoms in children may initially present without headache or fail to meet full diagnostic criteria. The observed evolution from initially unclassified presentations to migraine-related diagnoses supports the concept of pediatric vestibular migraine as a developmental disorder, in which the phenotypic expression may require longitudinal observation before diagnostic criteria are fully met. The introduction of the Bárány Society and International Headache Society consensus definitions for Vestibular Migraine of Childhood, probable Vestibular Migraine of Childhood, and Recurrent Vertigo of Childhood has provided a more robust framework for classification and reduced diagnostic ambiguity [19].

Recent comparative studies further demonstrate that these entities share overlapping yet distinguishable clinical profiles, supporting repeated clinical reassessment rather than early definitive labeling [10]. Our findings are consistent with this literature, indicating that migraine-related diagnoses are frequently revealed only through follow-up rather than at initial presentation.

4.5. Hemodynamic Etiologies

Hemodynamic mechanisms represent a clinically relevant and often under-recognized contributor to pediatric dizziness and vertigo. In our revised 2025 cohort, hemodynamic etiologies accounted for 16% (41/257), supporting the growing recognition that orthostatic intolerance syndromes and reflex syncope-related physiology can present predominantly as dizziness rather than frank syncope. This interpretation is consistent with contemporary pediatric autonomic literature, which describes orthostatic intolerance as common in children and adolescents, typically manifesting as dizziness, lightheadedness, fatigue, visual symptoms, and relief when supine [20]. Moreover, vasovagal syncope is widely reported as the most frequent cause of transient loss of consciousness in youth, accounting for the majority of pediatric syncope presentations—highlighting the high background prevalence of reflex/autonomic mechanisms in this age group [21]. In our cohort, tilt-table testing was performed in a small, clinically selected subgroup (13/257; 5%), and 62% (8/13) showed pathological results. While the sample is limited, this positivity rate is broadly in line with the high diagnostic yield reported in specialized pediatric syncope cohorts undergoing tilt testing, where positivity can exceed two-thirds depending on

referral selection and protocol; for example, a recent pediatric syncope series reported a 75% positive tilt-table response among those tested [22]. Therefore, our tilt-table findings are most plausibly interpreted as reflecting targeted testing in patients with higher pre-test probability, rather than population-level prevalence. Clinically, these data support a structured approach in which hemodynamic symptoms (orthostatic dizziness, presyncope, palpitations, visual dimming, symptom relief with recumbency) prompt focused autonomic evaluation, whereas indiscriminate tilt testing in unselected dizziness populations may be less informative [20]. Future prospective studies should include systematic autonomic evaluation protocols to allow more precise subclassification of hemodynamic causes in pediatric vertigo.

4.6. Neuroimaging in Pediatric Vertigo

Neuroimaging is commonly obtained in children presenting with vertigo or dizziness, particularly when neurological signs, persistent headache, or trauma are present; however, its diagnostic yield in the absence of specific clinical indicators is modest. In a tertiary referral cohort of children imaged for vertigo, abnormalities were more likely to correlate with neurological deficits or persistent headaches, and routine imaging without such red flags provided limited additional diagnostic value [23].

Consistent with this literature, in our 2025 revised cohort, cranial CT was performed in 160 children (62% of the total cohort) and brain MRI in 146 children (57%). Although CT scans were frequently obtained, pathological CT findings were rare (1%), reflecting a low yield in an unselected dizziness population. MRI demonstrated a higher frequency of abnormalities (17%) likely due to its greater soft-tissue resolution and ability to detect subtle central pathologies when clinically indicated. These observations align with pediatric imaging data showing that CT contributes less to etiological diagnosis in vestibular presentations compared with MRI when neuroimaging is selectively applied based on clinical suspicion [23].

Importantly, improved etiological classification through follow-up may reduce reliance on early extensive diagnostic testing and support a more rational, stepwise diagnostic strategy.

4.7. Psychogenic Vertigo

Age-related differences play an important role in the etiological spectrum of pediatric vertigo. In our original 2021 cohort, psychogenic vertigo was observed predominantly in older children and adolescents, whereas younger children more often presented with peripheral or benign episodic vestibular disorders. This age-dependent distribution suggested that psychological and functional contributors to vertigo become increasingly relevant with advancing age, a pattern consistently reported in the literature.

This observation is supported by population-based data showing that vertigo in children aged 3–17 years is significantly associated with emotional, behavioral, and concentration difficulties, with stronger associations in adolescents compared with younger children [24]. Clinical studies focusing on somatoform and psychogenic vertigo similarly report that these diagnoses are rare in early childhood but emerge more frequently during school age and adolescence, often in the context of psychosocial stressors and recurrent or persistent symptoms [25].

In the present 2025 revision, psychogenic etiologies accounted for 10% (26/257) of cases, a proportion comparable to that reported in other tertiary pediatric cohorts. Although the revision did not aim to reanalyze age-stratified outcomes, the persistence of psychogenic diagnoses across follow-up is consistent with our 2021 finding that psychological contributors are more likely to be identified in older children, particularly when

symptoms persist, and organic causes have been excluded. These findings underscore the importance of incorporating age-sensitive psychosocial assessment into the diagnostic pathway, especially during follow-up of adolescents with unexplained or recurrent vertigo.

4.8. Strengths and Limitations

4.8.1. Strengths

The principal strength of this study is the longitudinal re-evaluation of the same pediatric cohort using prospectively collected clinical data, combined with a retrospective revision of etiological classification after extended follow-up. This design enabled a direct within-cohort comparison between baseline diagnoses and those established after prolonged clinical observation. The relatively large sample size and the multidisciplinary diagnostic approach, involving pediatric neurology, otolaryngology, radiology, and ophthalmology, further enhance the robustness of the findings. Additionally, the application of updated diagnostic concepts and contemporary classifications of pediatric vestibular disorders increases the clinical relevance and translational value of the revised analysis.

4.8.2. Limitations

Several limitations should be considered when interpreting the results of this study. Although clinical data were collected prospectively, the etiological revision performed in 2025 represents a retrospective reassessment and therefore depends on the completeness and quality of available medical records. Not all children underwent follow-up evaluation: of the total cohort ($N = 257$), 52 children (20%) were not scheduled for follow-up based on clinical judgment at initial assessment, and 29 children (11%) did not attend the planned follow-up visit. Consequently, 32% of the cohort did not have a completed follow-up examination, which may have limited the ability to detect etiological changes in a subset of patients. In our cultural and healthcare context, non-attendance is often interpreted as reflecting symptom resolution and perceived clinical improvement; however, this assumption cannot be objectively verified, and alternative explanations cannot be excluded.

Furthermore, the duration of follow-up varied widely, ranging from 0 to 132 months, with a relatively short median. While extended follow-up enabled etiological reclassification in many children, shorter follow-up intervals in others may have reduced the likelihood of identifying diagnoses that emerge later in the clinical course.

Due to the absence of standardized follow-up intervals and systematically recorded time-to-reclassification data, we were unable to determine whether diagnostic refinement predominantly occurred within a specific early time window (e.g., within the first six months). Prospective studies incorporating structured reassessment timelines are needed to better define optimal follow-up intervals in pediatric vestibular disorders. The identification of baseline clinical predictors of diagnostic evolution would be of considerable clinical value. However, the present retrospective design did not allow formal predictive modeling. Future prospective cohort studies should aim to identify symptom patterns or clinical features associated with a higher likelihood of diagnostic reclassification.

Moreover, as this study was conducted at a single tertiary referral center, the findings may not be fully generalizable to primary or secondary care settings, where patient populations, referral patterns, and diagnostic resources may differ.

A further limitation concerns the absence of systematically recorded data on completion rates of specific vestibular tests and age-related differences in cooperation. Although repeat vestibular testing was performed in selected cases during follow-up and contributed to diagnostic refinement, detailed quantitative documentation of test repetition and age-

dependent feasibility was not available for retrospective analysis. This may have influenced diagnostic classification in some patients.

Finally, the hemodynamic category included cases of orthostatic intolerance and reflex-mediated mechanisms; however, detailed subclassification (e.g., formal differentiation between POTS, orthostatic hypotension, and vasovagal mechanisms) was not systematically documented, limiting further granularity of analysis.

5. Conclusions

This study demonstrates that pediatric vertigo and dizziness are frequently evolving conditions, in which etiological clarification often requires longitudinal follow-up. Revision of data after extended follow-up markedly reduced the proportion of unclassified cases and led to a redistribution toward central, hemodynamic, psychogenic, and peripheral vestibular diagnoses. These findings highlight that diagnostic uncertainty at initial presentation should be anticipated and does not imply the absence of an underlying disorder.

Overall, a longitudinal, multidisciplinary approach is essential to improve diagnostic accuracy, reduce unnecessary investigations, and optimize clinical management in children presenting with vertigo and dizziness.

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Abbreviations

The following abbreviations are used in this manuscript:

EEG	Electroencephalography
CT	Computerised tomography
MRI	Magnetic resonance imaging
BPPV	Benign paroxysmal positional vertigo
BPV	Benigne pediatric vertigo

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Article

Non-Instrumented DVA: Assessment of Performance and Clinical Feasibility in Children Ages 2 Through 13 Years

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Highlights

What are the main findings?

- Non-instrumented DVA testing is feasible for children aged 3 years and older and can be used to identify vestibular dysfunction across age groups.
- Performance on non-instrumented DVA testing does not differ significantly across age groups, with most children demonstrating results comparable to established adult normative values.

What are the implications of the main findings?

- The non-instrumented DVA can be administered to children using a simple methodology by newly trained clinicians and represents a promising and clinically feasible screening tool for identifying children with possible vestibular dysfunction. While most 3-year-olds can perform DVA testing, some may present challenges due to difficulty reading LEA Symbols or other behavioral factors.
- This tool shows promise as a feasible clinical option that may be integrated into pediatric physical therapy practice to assist in referral recommendations.

Abstract

Background/Objectives: Vestibular disorders can have functional consequences for children, including balance and gross motor delays, academic difficulties and behavioral manifestations; however, they are frequently undiagnosed in children. The purposes of this study were to evaluate the feasibility and clinical utility of performing a non-instrumented dynamic visual acuity (DVA) test as a primary screening tool for children and to examine typical performance on this non-instrumented DVA test in a large sample of children ages 2 through 13 years. **Methods:** A clinical DVA test was administered to a convenience sample of 208 children aged 2–13 years. Static visual acuity was assessed using a standard Snellen or LEA eye chart, depending on the child's ability to read letters. Dynamic visual acuity was then measured while the examiner manually rotated the child's head at 2 Hz (240 bpm). DVA was calculated as the number of lines of visual acuity lost with head movement. **Results:** All children aged 4 years and older and 67% of 3-year-olds successfully completed DVA testing. Most 2-year-olds and 33% of 3-year-olds were unable to complete DVA testing. Although the number of visual acuity lines lost with rotational head oscillations at 2 Hz varied between age groups, Kruskal–Wallis test indicated no significant difference in DVA scores between age groups ($K = 12.721$, $DF = 9$, $P = 0.176$).

Ninety percent of children who were able to perform DVA testing lost two or fewer lines of visual acuity with head rotations consistent with adult norms. **Conclusions:** This method of DVA testing is an easily accessible and promising clinically feasible screening tool for identifying children with vestibular dysfunction. The authors recommend widespread vestibular screening of children to facilitate rapid referral for diagnosis and treatment of children with vestibular dysfunction.

Keywords: dynamic visual acuity; DVA; vestibulo-ocular reflex; VOR; vestibular dysfunction; children; development; balance; academic performance; pediatric

1. Introduction

The vestibulo-ocular reflex (VOR) is essential for stabilizing gaze during head movements by enabling the eyes and head to move in equal and opposite directions. When the VOR is impaired, the eyes cannot adequately stabilize gaze during head motion, resulting in blurred vision and unstable fixation while moving. The dynamic visual acuity (DVA) test evaluates VOR function by measuring an individual's ability to see clearly while the head is in motion [1]. Poor performance on the DVA test may indicate VOR impairment consistent with underlying vestibular dysfunction, along with central problems such as cerebellar dysfunction [2].

Vestibular dysfunction in children is frequently under-recognized, thereby potentially contributing to delays in diagnosis. In part, this under-recognition stems from children's difficulty with articulating complex symptoms accompanied by vestibular disorders [3]. Unlike adults, children experiencing visual disturbances commonly associated with vestibular dysfunction may not raise complaints, as they are often unaware that a problem exists [4]. Additionally, symptoms may present as difficulty focusing during school or subtle balance and gross motor challenges, leading parents to assume the child is simply uncooperative or uncoordinated [3]. When subtle presentations occur alongside achievement of gross motor developmental milestones such as functional ambulation, clinicians may overlook the possibility of an underlying vestibular etiology [5]. Ultimately, delayed recognition and consequent late diagnosis of vestibular dysfunction in children can be attributed to the culmination of communication limitations, nonspecific symptom presentation, and developmental milestone attainment that masks underlying vestibular involvement. Despite estimates on the prevalence of pediatric vestibular disorders ranging from 0.7% to 15%, it has been shown that less than 20% of affected children are correctly diagnosed within one month of the onset of symptoms, and a large majority of affected children experience diagnostic delay exceeding one year [6].

Symptoms resulting from vestibular dysfunction can precipitate significant functional limitations, including increased risk of falls, impaired reading ability, and reduced participation in physical activities, with delayed diagnosis further complicating these issues [7–9]. Children with sensorineural hearing loss and concurrent vestibular impairment often show delays in motor skills and difficulty maintaining gaze stability, particularly when visual or somatosensory input is challenged, as well as deficits in sensory organization [4,9]. Without effective vestibular input during critical periods of childhood, typical postural control may not develop, leading to progressive motor delays. Vestibular dysfunction may also serve as a hidden barrier to learning. Franco and Panhoca [7] found a significant association between vestibular abnormalities and poor school performance and acknowledged that vestibular dysfunction can disrupt the sequential eye fixations required for reading, directly interfering with the development of fundamental academic skills. Moreover, com-

mon childhood conditions such as otitis media with effusion can have lasting vestibular consequences. Pazdro-Zastawny et al. [10] demonstrated that children with a history of middle ear effusion exhibited increased postural sway and nystagmus up to five years after treatment. Their findings underscore the potential for undetected dysfunction to persist and affect motor development. Beyond physical and academic consequences, pediatric vestibular disorders may impact psychosocial development. Children with vestibular disorders exhibit higher rates of emotional and behavioral difficulties, including anxiety, depression, and attention problems [11,12], and are at increased risk for cognitive and psychiatric comorbidities such as learning disabilities and developmental delays, highlighting the vestibular system's integral role in neurodevelopment [13]. Under-identified vestibular dysfunction leads to prolonged, untreated symptoms, compounding these challenges and diminishing social participation and quality of life [6]. Collectively, these findings highlight the urgent need for accessible, cost-effective screening tools to facilitate early identification and intervention.

Several advanced diagnostic tools exist for evaluating vestibular dysfunction in children, including Videonystagmography (VNG), Rotary Chair Testing, Video Head Impulse Test (vHIT), and Vestibular Evoked Myogenic Potentials (VEMPs). While these instrumented tests provide comprehensive diagnostic data, they are costly and require specialized equipment and advanced clinician training. Consequently, such resources are often unavailable and inaccessible in many pediatric clinical settings [6,14,15]. Thus, these tests may be impractical as first-line screening tools, and vestibular dysfunction frequently goes undetected or is diagnosed late, contributing to prolonged symptoms and developmental consequences. Barriers such as limited time for screening have also been reported by pediatric rehabilitation professionals, further reducing the utilization of vestibular assessments as screening tools within clinical practice [15,16]. This gap underscores the need for accessible, efficient screening methods to identify children who warrant referrals for comprehensive vestibular evaluation. In response to these barriers, standardized clinical balance tests such as the Pediatric Balance Scale (PBS), pediatric Modified Clinical Test of Sensory Interaction in Balance (mCTSIB) and Timed Up and Go (TUG) have been recommended as low-cost alternatives for assessing functional balance in children [15]. However, these tools primarily evaluate functional balance and postural control rather than directly assessing vestibular system function, thereby potentially limiting comprehensive evaluation critical for pediatric patients with balance deficits [17]. Ultimately, vestibular function screening is underutilized within pediatric settings, resulting in missed opportunities for early detection and intervention [17]. The non-instrumented DVA presents an accessible and practical screening tool for vestibular system function that enables clinicians to quickly and accurately rule out vestibular dysfunction. Broader adoption of this screening tool could improve the timely identification of vestibular pathology and optimize developmental outcomes.

Among available vestibular tests, the DVA test is a clinically relevant option for assessing vestibular function in adults, demonstrating strong diagnostic validity and reliability for the instrumented version and promising results for the non-instrumented version. Herdman et al. [1] established the instrumented (computerized) version of DVA testing as a highly reliable test for assessing VOR function in adults as it ensured head velocities were kept within a range that isolated VOR from other eye-tracking systems. Testing ultimately demonstrated high sensitivity and specificity when differentiating between adults with vestibular deficits versus those in the control group [1].

Given the excellent reliability and diagnostic validity of the instrumented version of DVA testing, subsequent research on non-instrumented DVA testing has been conducted to determine the utility of DVA testing that accommodates more accessible and less ex-

pensive methods [18–21]. Early research from Longridge and Mallinson [18] established strong predictive parallels between the “Dynamic Illegible E” test (DIE), a version of non-instrumented DVA testing, and the instrumented caloric testing, a gold standard diagnostic test for unilateral vestibular hypofunction. The authors ultimately reported a positive correlation ($p < 0.05$) between the two tests as the degree of abnormality within DIE testing increased with the degree of caloric reduction. Because all control group participants lost no more than one line on the DIE test, the authors defined a DIE cutoff score as a loss of more than two lines. Furthermore, Roberts and Gans [19] reported non-instrumented horizontal-plane DVA testing as more sensitive towards VOR impairments than non-instrumented vertical-plane testing, with an overall moderately high negative predictive value of 83.6%. Recent studies report the “bedside” applicability of non-instrumented DVA testing for detecting VOR impairment in adults [21] and its high clinical feasibility, as the test is fast, inexpensive, and accessible [20]. Overall, while variability in testing protocol has limited the consistency of reported reliability and validity measures, the non-instrumented DVA test has been widely recognized as a clinically feasible screening tool for identifying adults with possible vestibular dysfunction.

Although research remains limited, studies indicate that DVA testing serves as a clinically feasible method for assessing vestibular function in children. Early work by Rine introduced a simple, non-instrumented DVA test using LEA symbols, demonstrating excellent test–retest reliability ($ICC = 0.94$) and 100% sensitivity and specificity for detecting bilateral vestibular hypofunction [4]. This study confirmed DVA testing as appropriate for children as young as three years old. Subsequent research reinforced the clinical effectiveness of DVA testing, reporting high sensitivity for identifying vestibular hypofunction in children and establishing minimal detectable change scores (8 optotypes), which enable clinicians to monitor meaningful progress over time [22]. The non-instrumented DVA test for children is both cost-effective and time-efficient, requiring only a vision chart, a chair, and a metronome, compared to more involved and costly diagnostic tests. It also demonstrated strong diagnostic accuracy with a negative likelihood ratio of 0.18 for ruling out vestibular hypofunction [22], thereby allowing clinicians to quickly and accurately rule out vestibular hypofunction. Together, these findings provide strong evidence for the construct validity and reliability of non-instrumented DVA testing as an accessible, cost-effective screening tool that can guide referral decisions and complement diagnostic evaluations in pediatric settings.

Adult norms for normal DVA performance, clinically defined as fewer than two lines of visual acuity loss [18], have been reported in the literature. Using the computerized (instrumented) NIH Toolbox DVA (cDVA) test, Li et al. conducted a large ($n = 3992$) cross-sectional study across ages 3–85 years and established normative reference values [23]. Results showed a median cDVA difference of 0.070–0.080 logMAR (≈ 1 line of visual acuity loss during head movement) for adults ages 18–49. In pediatric populations, Rine and Braswell demonstrated through use of a non-instrumented clinical approach that children as young as three years typically show two or fewer lines of visual acuity loss during head movement, which is considered within normal limits [4]. The discrepancy in reporting metrics, logMAR difference versus lines of visual acuity lost, highlights a lack of standardization in normative data, indicating a need for further research. Despite this variability, normative adult DVA benchmarks are clearly defined in the literature, helping to serve as a reference for DVA performance interpretation in other populations.

Although comprehensive diagnostic tests are valuable, their cost, instrumentation, and specialized training requirements make the tests hard to access and impractical as first-line screening in routine pediatric care. In contrast, non-instrumented DVA testing directly analyzes VOR function with favorable reliability, sensitivity, and practical feasibility in

children [4,22]. Despite its promising applicability in pediatric settings, research on the usage of the non-instrumented DVA as a pediatric screening tool is limited, underscoring a need for further investigation of its utility, cost-effectiveness, and overall practicality in pediatric populations. The purposes of this study were to evaluate the feasibility and clinical utility of performing a non-instrumented DVA test as a primary screening tool for children and to examine typical performance on this non-instrumented DVA test in a large sample of children ages 2 through 13 years.

2. Materials and Methods

2.1. Participants

A convenience sample of 208 children (91 males and 117 females) aged 2 through 13 years was recruited from schools, churches, preschools, and other community organizations. All children were developing typically per parent report. Study inclusion criteria included the ability to follow simple verbal instructions and consistently identify either shapes on the LEA symbols chart (Good-Lite Company, Elgin, IL, USA) or letters on the Snellen eye chart (PECULA, Shenzhen, China). Children were excluded from study participation if parents reported any neurologic or developmental health conditions, seizures within the previous 6 months, or any other physician-recommended activity limitations that would prohibit safe participation in the study.

2.2. Rater Training

Twenty-nine raters (27 entry-level physical therapy students and 2 physical therapy faculty) participated in data collection. All raters received formal training in the administration and scoring of the DVA by two Belmont University Physical Therapy Faculty members, both of whom are board-certified clinical specialists in Neurologic Physical Therapy. Each student rater completed a training course that included demonstration and guided practice in administering and scoring the DVA with peers and non-participant children. All data collection occurred under the direct supervision of two faculty mentors.

2.3. Materials and Equipment

This study utilized a non-instrumented version of the Dynamic Visual Acuity (DVA) test [4,24]. Visual acuity was assessed using a Snellen eye chart [25] for children who were able to identify letters and a LEA symbol chart [26] for children unable to read letters. Equipment required for DVA testing included a chair with back support and appropriate height to ensure the child's feet were supported flat on the floor, along with a tape measure, an eye chart, and a metronome.

2.4. Procedures

Prior to participation, each parent completed written informed consent and a health history form on behalf of their child. Participants aged 7 to 13 years also provided written assent using an age-appropriate child assent form. Parents signed a waiver of assent for children aged 6 and under. A preschool-level script describing the study was read to these young children before they were asked for verbal assent. All procedures and consent forms were approved by the Belmont University Institutional Review Board (IRB #1281). After obtaining informed consent and age-appropriate assent, each participant was assigned an identification number to ensure confidentiality throughout testing and data management.

The testing protocol was standardized across all sites to minimize variability. All testing was conducted individually, with two examiners assisting with testing each child. Faculty mentors were also present during testing. The investigators confirmed the date of birth, absence of exclusion criteria, and the child's ability to follow simple instructions

with the parent before initiating testing. Two children were excluded from the study. One child had remarkable atypical oculomotor function and was subsequently referred to her pediatrician with a recommendation for neuro-ophthalmology evaluation. Visual acuity could not be assessed in the other child because she did not bring her glasses and could not read the Snellen chart without them.

Test administrators then confirmed each child's ability to identify letters or shapes to determine the appropriate eye chart. If children could not quickly and easily read letters, then the LEA symbols chart was used, and if they could not consistently identify letters or LEA symbol shapes, they were disqualified from participation. A reference card displaying the LEA symbols was used prior to testing to ensure the participant could identify each symbol. Children were not disqualified if they were consistent in how they mislabeled LEA symbols shapes. For example, many children consistently mislabeled the "apple" as a heart or the "house" as a triangle. Participants who normally wore glasses or contact lenses wore them during testing.

Each child was positioned in an appropriately sized chair with a back that enabled the child to sit with their back supported and their hips and knees at 90 degrees of flexion with their feet flat on the floor. The chair was placed at the appropriate distance from the eye chart per protocol (10 feet from the LEA symbols chart, 20 feet from the Snellen chart) with the chart affixed to the wall at the child's eye level. Testing was conducted individually with two testers assisting with DVA testing. One rater (R1) stood behind the child, and a second rater (R2) was positioned by the eye chart.

Binocular static visual acuity was assessed first and recorded in logMAR units, with the lowest line read with two or fewer errors documented. Two trials of dynamic visual acuity with head rotations were then performed. R1 positioned the child's head in 30 degrees of cervical spine flexion and passively rotated their head horizontally right and left 20–30 degrees (approximately 15 degrees to each side) in a sinusoidal pattern at a frequency of 2 Hz, guided by a metronome set to 240 beats per minute. Head rotation at 2 Hz was selected based on previously published pediatric and adult DVA protocols demonstrating that this frequency provides adequate VOR challenge while remaining safe. Consistency was supported through structured rater training, faculty oversight, and metronome pacing, though some amplitude variability inherent in manual methods is acknowledged [8]. R2 was positioned next to the eye chart and used a piece of white paper to cover the lines immediately below the line being read by the child to help maintain visual focus. R2 also verified accuracy of the child's responses as they read each line on the eye chart. The lowest line the participant could read, with two or fewer errors documented during head movement, was recorded. The number of lines lost between static and dynamic conditions was calculated and documented as the DVA score. Each child completed one practice session followed by two test trials. A small number of children refused a second trial of DVA with head movement.

3. Results

Children were divided into 12 groups in one-year age increments for descriptive data analyses. The 12- and 13-year-old groups were subsequently combined for remaining statistical analyses due to relatively small numbers in each group (7 and 5, respectively). Number of lines of visual acuity lost with head rotations was used for all data analyses. Of the 208 children, 185 were able to successfully complete DVA testing (Table 1). Only one of the 15 two-year-olds was able to complete DVA testing; results for this child were not used for further data analysis. Two-thirds of 3-year-olds and all children four years and older were able to successfully complete DVA testing. Although mean lines lost with head rotations differed slightly between age groups (Figure 1), a Kruskal–Wallis test indicated

no significant difference in DVA scores between age groups ($K = 12.721$, $DF = 9$, $P = 0.176$). Ninety percent of children who were able to perform DVA testing lost two or fewer lines of visual acuity with head rotations consistent with adult norms [23] (Figure 2).

Table 1. Identifies numbers and percentages of children successful with DVA testing and descriptive statistics for this group.

Age (years)	Total Number	Children Successful with DVA Testing		Lines of Visual Acuity Lost with Head Rotation			
		n	%	Mean	SD	95% CI Lower Bound	95% CI Upper Bound
2	15	1	6.7				
3	28	19	67.9	1.21	0.23	0.75	1.67
4	29	29	100	1.35	0.19	0.97	1.72
5	31	31	100	1.29	0.18	0.93	1.65
6	19	19	100	1.00	0.24	0.53	1.47
7	16	16	100	0.53	0.25	0.04	1.02
8	19	19	100	0.84	0.23	0.38	1.30
9	12	12	100	0.92	0.29	0.34	1.50
10	15	15	100	0.80	0.26	0.28	1.32
11	12	12	100	0.75	0.29	0.17	1.33
12&13	12	12	100	1.17	0.29	0.59	1.75

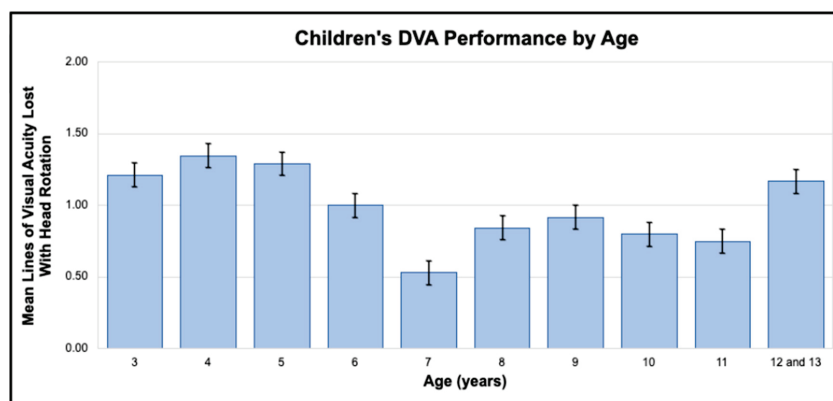


Figure 1. Means and standard deviations for lines of visual acuity lost with head rotation at 2 Hz.

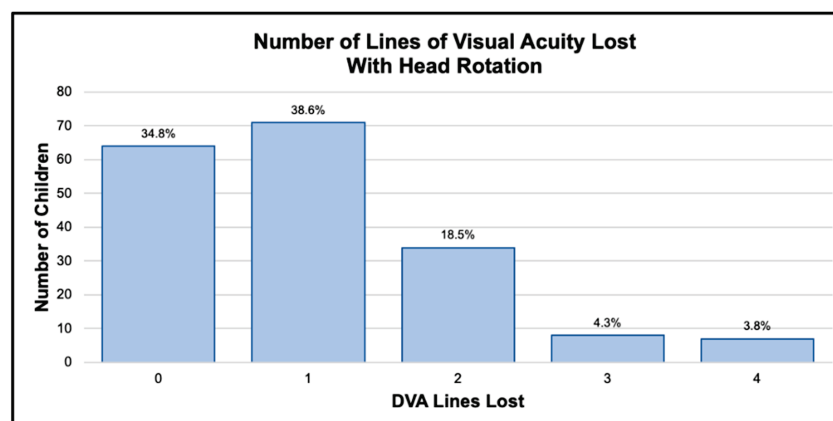


Figure 2. Numbers and percentages of children obtaining each score on DVA testing.

4. Discussion

The purpose of this study was to evaluate the clinical utility of the non-instrumented Dynamic Visual Acuity (DVA) test as a primary screening tool for vestibular dysfunction in pediatric populations and to examine the typical DVA performance in a large sample of children aged 2 to 13 years. The results of this study indicate that the non-instrumented DVA can be administered to children using a simple methodology by newly trained clinicians and yields outcomes comparable to those reported in both instrumented [23] and non-instrumented DVA studies [4].

All children aged 4 and older completed DVA testing. Two-year-old children were largely unable to complete the test, indicating limited feasibility in this age group. All but one two-year-old and one-third of three-year-olds were unable to complete DVA testing. Although two-thirds of 3-year-olds were able to complete DVA testing, many required more time and patience from the examiner to complete the test. The most common limitation to DVA testing in these children was the inability to consistently identify the LEA symbols. Children were not required to accurately name the LEA symbols but had to be consistent with the names they chose for the symbols. For example, some young children referred to the “apple” symbol as a “heart” and the “square” symbol as a “triangle.” Some two- and three-year-olds also refused to let the examiner shake their head during the dynamic portion of DVA testing. Comparisons across studies are difficult due to variations in protocol and scoring.

No significant differences in DVA performance were found across age groups, suggesting consistency across childhood. Most children (92%) in this study, who successfully completed DVA testing, lost two or fewer lines of visual acuity, which is consistent with previously reported adult norms on instrumented [23] and non-instrumented clinical DVA [18]. Health histories of the 15 children who successfully completed DVA testing but lost 3 or more lines of visual acuity ($DVA \geq 3$) with head rotations were further examined for commonalities and differences between this group and the remaining 170 children who lost two or fewer lines of visual acuity ($DVA \leq 2$). Twelve parents reported health conditions that could be potentially associated with atypical vestibular function or balance (Table 2). The most common of these reported conditions was ear infections. Adverse effects of ear infections on vestibular function and balance in children have been well documented in the literature [27–29]. Thirty-three percent of children in the $DVA \geq 3$ group had ear infections in the year prior to testing, and 67% had a lifetime history of ear infections. Rates of ear infections for the $DVA \leq 2$ group were 38% in the previous year, with 53.5% of children in this group having a lifetime ear infection history. While it appears that children in the $DVA \geq 3$ group may have a higher lifetime rate of ear infections, all information about ear infection history was provided via parent report. Parents were asked to report how many ear infections their child had in the past year and whether the child had experienced any ear infections before that period. They were not asked about ear infection effusion or severity which limits comparison of DVA performance in children with and without ear infection history. This examination of health history in the $DVA \geq 3$ group should be considered exploratory based on the following factors: (1) small sample size, (2) health history was obtained by parent report and (3) vestibular dysfunction was not confirmed with instrumented testing. Future studies should include prospective examination of DVA performance in children who have current documented ear infections with follow-up DVA testing to examine potential long-term effects of ear infections on vestibular function. Finally, the authors recommend consideration of targeted DVA testing for children with ear infection history both in clinical settings and in future studies where vestibular dysfunction may influence study outcomes.

Table 2. Provides detailed information for children who lost 3 or more lines of visual acuity.

Child Number	Age	Lines Lost	Ear Infections in Last Year	Evidence of Ear Infections in Last Year or Prior	Parent Comments
1	3	4	yes	yes	Skull fracture, feeding therapy, reflux, 2 sets of ear tubes
2	3	3	no	no	
3	4	4	no	no	Resolved gross motor delays
4	4	4	yes	yes	Recurring ear infections, ear tubes
5	4	3	yes	yes	2 sets of ear tubes
6	4	3	no	no	
7	5	4	no	no	Behavioral health, speech therapy
8	5	4	no	no	
9	5	3	yes	yes	ADHD
10	5	3	no	yes	
11	5	3	no	yes	Ear tubes at 1.5 years old
12	6	3	no	yes	
13	8	4	yes	yes	
14	8	3	no	yes	
15	10	4	no	yes	ADHD, motion sickness
Total			5	10	

Two children aged 5 years and older (22%) in the $DVA \geq 3$ group had parent-reported Attention Deficit/Hyperactivity Disorder (ADHD), which was very similar to the parent-reported rate (20%) in the $DVA \leq 2$ group (Table 2). The authors chose to only include children ages 5 years and older in these calculations, as ADHD is often not diagnosed until school age. Although the relationships between ADHD have not been examined extensively, Gur-Hartman et al. found that children with diagnosed ADHD demonstrated evidence of vestibular hypofunction on DVA testing using a method very similar to that of the current study [24]. Children with ADHD also had reduced balance ability compared to peers without ADHD as measured by 9 items on the Bruininks–Oseretsky Test of Motor Proficiency (BOT-2). These differences were even more pronounced in children with ADHD and a comorbid diagnosis of developmental coordination disorder (DCD). Although the current study demonstrated similar rates of ADHD in children with typical and atypical DVA results, it should be noted that the sample sizes were small. Additionally, all information about ADHD diagnoses was provided via parent report and not corroborated with diagnostic testing.

The non-instrumented DVA test serves as a highly feasible and practical screening tool for pediatric populations, primarily due to its minimal equipment requirements and low cost. Rine & Braswell [4] and Christy et al. [22] described DVA testing methods for clinical settings. Both found clinical DVA testing to be reliable and able to accurately identify children with vestibular dysfunction. Unlike advanced diagnostic measures such as Videonystagmography or Rotary Chair Testing, which are often inaccessible in pediatric settings, the non-instrumented DVA requires only a LEA and/or Snellen vision chart, a chair, and a metronome. The simplicity of the equipment required allows the test to be successfully administered across a wide variety of environments; in this study, the DVA

was administered in therapy gyms, daycare centers, gymnastics facilities, churches, and private homes. DVA testing requires less than 10 min per child to administer, overcoming a major barrier in pediatric care, where clinicians may bypass vestibular screening due to schedule constraints [30]. By providing accurate results in diverse settings without the need for expensive, advanced technology, the non-instrumented DVA offers an efficient solution for identifying children with vestibular hypofunction [4]. The current study successfully utilized a similar method to that first described by Rine & Braswell [4] and more recently by Gur-Hartman [24] to assess DVA in children aged 3 through 13 years. It is unique in that entry-level physical therapy students administered the DVA to a large sample of children, following a structured training protocol. This indicates that novice clinicians can become proficient at DVA administration with minimal training.

The non-instrumented DVA uniquely contributes to pediatric vestibular screening because it directly assesses an individual's ability to use VOR to maintain visual stability during head movement without specialized equipment or training; the DVA test additionally provides diagnostic information not captured by functional balance testing. By offering a rapid way to identify visual-vestibular deficits, the DVA can guide timely referral and facilitate earlier initiation of targeted vestibular rehabilitation, ultimately supporting improved functional outcomes.

A primary limitation of non-instrumented DVA testing relates to variability across methodologies, which complicates direct comparison of results. Although standardized head movements at approximately 2 Hz are a shared requirement, studies differ in optotype choice, scoring approach, and termination criteria. Also of note is the possible head movement variability of non-instrumented DVA testing. Despite these inconsistencies, research consistently demonstrates that reduced DVA performance is associated with underlying VOR impairment [4,18,24]. The simplified protocol in this study (i.e., minimal equipment, short testing duration, high feasibility for novice and experienced clinicians alike) enhances clinical practicality but may reduce sensitivity relative to instrumented assessments, particularly for detecting subtle changes over time. Future investigators may wish to add head-mounted sensors to monitor head amplitude variability. While this may improve accuracy, it will add to the cost, which may be prohibitive and decrease feasibility in many clinics. Nevertheless, its efficiency and diagnostic relevance reinforce its value as an efficient first-line screen to identify children who may benefit from further vestibular rehabilitation.

5. Conclusions

Non-instrumented DVA testing is a clinically feasible and accessible screening tool for children aged 3 years and older, with performance levels that demonstrate consistency across age groups and align with established adult norms [23]. Its minimal equipment requirements and the ability for novice clinicians to achieve proficiency make it an efficient first-line assessment suitable for diverse pediatric clinical settings. Implementing screening can facilitate earlier identification of vestibular dysfunction, particularly in children with contributing health histories like ear infections, thereby guiding timely referrals for comprehensive evaluation [17]. Widespread adoption of the non-instrumented DVA test has the potential to optimize functional and developmental outcomes through the initiation of targeted vestibular rehabilitation [5,24].

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Informed Consent Statement: Informed consent was obtained from parents or guardians of all children participating in the study, and age-appropriate assent was obtained from each participant.

Data Availability Statement: Data are unavailable to others due to privacy restrictions enforced by the United States government (HIPAA, FERPA). The protocol for this study approved by Belmont University Institutional Review Board does not include permission for data sharing. The authors are willing to collaborate with others who would find this research valuable.

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Article

ABR Features in Ski-Slope Hearing Loss for Hearing Threshold Estimation: A Comparative Clinical Study of Click and CE-Chirp Stimuli

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Abstract

Background: Auditory brainstem responses (ABRs) are widely used for objective hearing threshold estimation in both adults and children. Click and CE-Chirp stimuli differ substantially in cochlear activation and neural synchrony, yet their relative performance in patients with ski-sloping hearing loss remains insufficiently characterized, particularly with regard to pediatric diagnostic implications. **Methods:** This study compared ABRs elicited by click and CE-Chirp stimuli in adults with ski-sloping sensorineural hearing loss. The same comparison was also performed in a pediatric cohort including hearing-impaired and normal-hearing children. Adult subjects were further stratified according to audiometric configuration (DROP 1 kHz vs. DROP 2 kHz). ABR thresholds, wave V latency, amplitude, and detectability were analyzed across stimulus types and intensity levels. Associations between ABR thresholds and behavioral audiometric measures were also examined. **Results:** In adults with ski-sloping hearing loss, CE-Chirp stimulation yielded significantly lower ABR threshold estimates than click stimulation, particularly in the DROP 2 kHz subgroup, and showed stronger correlations with behavioral pure-tone averages across low-, mid-, and high-frequency ranges. Wave V latencies were consistently shorter with CE-Chirp stimulation, while wave V amplitudes did not differ significantly between stimuli at suprathreshold levels. In children, ABR thresholds obtained with CE-Chirp were generally equal to or lower than those obtained with clicks, although statistical significance was limited by sample size. CE-Chirp stimulation was associated with shorter wave V latencies in both hearing-impaired and normal-hearing children and produced larger wave V amplitudes at selected suprathreshold intensities in hearing-impaired children. **Conclusions:** Click and CE-Chirp stimuli provide complementary information in ABR assessment. While click stimulation remains essential for robust waveform identification, CE-Chirp stimulation appears to offer advantages in threshold estimation and neural synchrony, particularly in ski-sloping hearing loss and pediatric evaluations. Discrepancies between click- and CE-Chirp-derived ABR thresholds should not be attributed solely to maturational or synchrony-related factors but may warrant further frequency-specific audiological assessment to optimize diagnosis and rehabilitation strategies.

Keywords: auditory brainstem response; click; CE-Chirp; ski-slope hearing loss; pediatric audiology; high-frequency hearing loss

1. Introduction

Auditory brainstem responses (ABRs) play a central role in objective hearing assessment across the lifespan and are particularly critical in pediatric populations, where reliable behavioral audiometry is often not feasible. ABRs are widely used for newborn hearing screening, diagnostic threshold estimation, and longitudinal monitoring of auditory function. Among the factors influencing ABR outcomes, stimulus type represents a key determinant, as different stimuli engage distinct cochlear regions and neural synchrony mechanisms, in accordance with the tonotopic organization of the cochlea [1].

Click stimuli have long been considered the clinical standard for ABR recordings, providing robust and well-defined waveforms through broadband cochlear activation. However, due to the tonotopic organization of the cochlea and cochlear travel-time delays, click-evoked responses predominantly reflect neural activity arising from the mid-to-high frequency regions, typically between 2 and 4 kHz [2–4]. This intracochlear dispersion reduces neural synchrony and may result in decreased response amplitude and less accurate threshold estimation, particularly in specific audiometric configurations [5].

To overcome these limitations, CE-Chirp stimuli were developed to compensate for cochlear dispersion by introducing frequency-specific temporal delays, thereby enhancing neural synchrony across a broader cochlear region [3,6]. The CE-Chirp stimulus, characterized by a flat amplitude spectrum and wide frequency coverage, has been shown to elicit larger response amplitudes and improved waveform morphology compared to click stimuli, especially at low stimulus [7–9]. These properties have led to widespread adoption of CE-Chirp stimulation in pediatric audiology and neonatal hearing assessment [10]. However, it is important to note that CE-Chirp stimuli were developed based on latency models derived from normally functioning cochleae, assuming typical frequency-place mapping and normal cochlear tuning [11].

Ski-sloping hearing loss, characterized by relatively preserved low-frequency thresholds and a steep decline at higher frequencies [12,13], represents a particularly challenging condition for ABR-based threshold estimation. In pediatric populations, high-frequency hearing loss may negatively affect speech perception and language development, making early and accurate diagnosis essential [14–16]. Importantly, several studies have demonstrated that timely audiological intervention, including early hearing amplification or cochlear implantation when indicated, leads to significant improvements in speech perception and language development in children [17,18]. These findings further emphasize the clinical significance of performing ABR assessments with both click- and CE-Chirp stimuli in a complementary manner to optimize threshold estimation and facilitate early intervention where appropriate. In this context, broadband click stimulation, which predominantly activates basal cochlear regions, may misrepresent auditory thresholds in the presence of steep high-frequency losses [19]. Although CE-Chirp stimulation may provide improved neural synchrony, its diagnostic performance in ski-sloping hearing loss remains incompletely understood, particularly in children.

While previous studies have compared click and CE-Chirp stimuli in normal-hearing individuals and in generalized sensorineural hearing loss [1,11], data focusing specifically on ski-sloping hearing loss are limited. This gap is especially relevant in pediatric clinical practice, where stimulus selection may influence diagnostic interpretation and subsequent rehabilitative decisions.

The present study aims to compare ABRs elicited by click and CE-Chirp stimuli in adults with ski-sloping hearing loss and in a pediatric cohort, with particular attention to threshold estimation, waveform morphology, and stimulus-related discrepancies. The adult cohort provides a reference model in which ABR findings can be interpreted alongside behavioral audiometry, whereas the pediatric cohort represents the primary clinical target,

where objective electrophysiological measures are most critically needed. By stratifying adult subjects according to audiometric configuration, this study seeks to provide insights that may inform the interpretation of ABR findings in pediatric settings when ski-sloping hearing loss is suspected.

2. Materials and Methods

2.1. Ethical Approval Section

This study was conducted in accordance with the principles of the Declaration of Helsinki. Data were examined in compliance with Italian privacy and sensitive data laws and the internal rules of the Padova University Otolaryngology Section. All patients or their parents signed a consent form for privacy disclosure in managing personal data for clinical and scientific purposes.

The present study was approved by the Ethics Committee of the Azienda Ospedale Università Padova (protocol No. AOP2444).

2.2. Participants

The study cohort included 32 participants: 24 adults (mean age: 45.7 ± 21.2 years) and 8 children (mean age: 3.1 ± 0.83 months). Adults comprised 14 subjects with ski-slope hearing loss and 10 normal-hearing controls; all patients had a sensorineural ski-slope audiometric configuration. Two subgroups were defined based on the audiometric configuration: (i) subjects with normal thresholds up to 1 kHz followed by a sharp threshold drop at higher frequencies (DROP 1 kHz group) and (ii) subjects with preserved thresholds up to 2 kHz followed by a sharp threshold drop from 4 kHz onward (DROP 2 kHz group).

This stratification (i.e., subdivision in DROP 1 kHz group and DROP 2 kHz group) was based on the hypothesis that the onset frequency of the steep high-frequency decline may differentially influence click- and CE-Chirp-evoked ABR features and threshold estimates due to stimulus-dependent weighting of cochlear contributions.

A control group of normal-hearing subjects was included for waveform characterization analyses. Adult and pediatric cohorts were analysed separately. Normal-hearing adult controls were included for detailed waveform characterization in adults, allowing comparison of ABR morphology and response features with the hearing-impaired adult cohort. Pediatric controls were used exclusively for comparative analyses within the pediatric cohort, due to age-dependent differences in ABR latencies and amplitudes.

The pediatric group included 7 with hearing loss and 7 normal-hearing controls. Participants were rigorously screened to exclude middle ear pathologies or neurological impairments. For pediatric participants, cases with obvious complications of otitis media, syndromic hearing loss associated with structural abnormalities of the middle or inner ear, or other known congenital malformations were explicitly excluded. The term “neurological impairments” was used to refer specifically to clinically diagnosed central nervous system disorders or developmental conditions that could affect auditory processing and ABR reliability, such as cerebral palsy, severe developmental delay, or documented central auditory pathway anomalies. Children with these conditions were not included in the study. These criteria ensured that the pediatric cohort comprised children with isolated sensorineural hearing loss.

2.3. Audiometric Assessment

For adults, audiometric evaluations encompassed otoscopy, pure-tone audiometry, speech audiometry, and the Italian Matrix Test (IMT) for speech-in-noise perception. Initially, otoscopy was performed to rule out the presence of impacted earwax or evident pathologies of the tympanic membrane.

Pure-tone audiometry was conducted using the clinical audiometer Madsen Astera2 (Natus Sensory Inc., Schaumburg, IL, USA) in a soundproofed booth. Air conduction thresholds were measured using TDH39 headphones, presenting pure tones at frequencies of 250, 500, 750, 1000, 2000, 3000, 4000, 6000, and 8000 Hz for each ear separately. Bone conduction thresholds were assessed using a bone vibrator placed on the mastoid at frequencies ranging from 500 to 4000 Hz. Contralateral masking was applied when required using narrow-band noise (NBN), calibrated according to Studebaker's method [20].

To quantitatively describe audiometric configuration, a slope index was derived as a continuous measure reflecting the steepness of the high-frequency threshold decline relative to preserved low-frequency hearing. The slope index was calculated as the difference between average high-frequency and low-frequency pure-tone thresholds, adapting the frequency ranges to the two audiometric subgroups. In the DROP 2 kHz group, the index was computed as the mean threshold at 4 and 8 kHz minus the mean threshold at 0.5, 1, and 2 kHz. In the DROP 1 kHz group, the index was computed as the mean threshold at 2, 4, and 8 kHz minus the mean threshold at 0.5 and 1 kHz. Higher values indicate a steeper ski-slope configuration.

For children, assessments included otoscopy, tympanometry, distortion product otoacoustic emissions (DPOAE), and ABRs to establish hearing thresholds.

2.4. ABR Protocol

ABRs were recorded using the Neuro-Audio system Version 1.0.104.1 (Inventis S.r.l., Padua, Italy) using standard surface electrode montage. The nHL values used in this study were based on the calibration provided by the ABR system manufacturer. Click and CE-Chirp stimuli were presented at 80, 60, 40, and 20 dB nHL using alternating polarity. Analysis windows were set at 10 ms for Click stimuli and 15 ms for CE-Chirp stimuli, with a stimulation rate of 21 Hz. Up to 2000 sweeps per stimulus were averaged, with a ± 10 mV rejection threshold and a recording noise level (RNL) set at 90 nV as an index of recording quality. Electrode impedance was kept below 3 k Ω and monitored per channel. Signal quality was optimized using adjustable high-pass (0.01–3000 Hz) and low-pass (10–10,000 Hz) filters, with an optional 50/60 Hz notch filter.

For each stimulus and intensity, the presence or absence of wave V was determined, and wave V latency and amplitude were measured when identifiable. The ABR threshold was defined as the lowest stimulus intensity at which a replicable wave V response was identified.

2.5. Quality Assessment of ABR Exams

To assess the clinical utility of each stimulus, 10 experienced ABR clinicians were asked to select which trace (click or CE-Chirp) they would use to define the patient's threshold. ABR recordings from 5 patients were randomly selected, and traces were displayed as images with all identifying information removed to prevent recognition of the stimulus type.

To minimize subjective variability in threshold determination, clinicians apply predefined objective criteria. Specifically, a response was considered present only when waveform components demonstrated high replicability across repeated averages and when peak amplitudes clearly exceeded the estimated residual background noise level (typically $\geq 3:1$ signal-to-noise ratio) [21,22].

2.6. Statistical Analysis—Adult Group

Statistical analyses were performed using jamovi (version 2.6.44). Descriptive statistics are reported as mean $\pm$ standard deviation or median and interquartile range, as appropriate. Paired comparisons between click- and CE-Chirp-derived ABR thresholds

were performed using the non-parametric Wilcoxon signed-rank test, both in the overall hearing-impaired cohort and within audiometric subgroups. The probability of detecting wave V at a given intensity level was analyzed using mixed-effects logistic regression models (binomial distribution, logit link). Stimulus type, intensity, audiometric group, and RNL were included as fixed effects, with subject included as a random intercept to account for repeated measures across intensities and stimuli. Wave V amplitude was analyzed using linear mixed-effects models. Amplitude values were log-transformed to improve distributional properties. Fixed effects included stimulus type, intensity, audiometric group, and RNL, with the subject modeled as a random intercept. Sensitivity analyses were performed by restricting the analysis to suprathreshold levels (40–80 dB nHL) and by testing stimulus-by-intensity interactions. Wave V latency was analyzed using linear mixed-effects models restricted to suprathreshold intensities (60 and 80 dB nHL) to ensure stable waveform identification. Fixed effects included stimulus type, intensity, audiometric group, and slope index, with subject included as a random intercept. Interaction terms were explored in secondary analyses. Given the limited sample size within audiometric subgroups, subgroup-related analyses (including subgroup terms in mixed-effects models and subgroup-stratified comparisons) were considered exploratory and were interpreted cautiously, focusing on effect direction and uncertainty rather than definitive subgroup inference. Associations between ABR thresholds and behavioral audiometric measures were assessed using Spearman's rank correlation coefficient. Given the ordinal nature of ABR threshold data and the presence of tied values, Kendall's tau-b was also calculated as a robustness measure.

2.7. Statistical Analysis—Pediatric Group

Given the small pediatric sample size, non-parametric statistical tests were adopted, and analyses in this cohort were considered exploratory in nature. Analyses were conducted separately in hearing-impaired children and normal-hearing controls when appropriate. ABR thresholds obtained with click and CE-Chirp stimuli were compared in hearing-impaired children using the Wilcoxon signed-rank test. Differences in wave V detectability at low stimulus intensity (40 dB nHL) were assessed using McNemar's test for paired binary data. Wave V latency and amplitude were analysed at suprathreshold levels (60 and 80 dB nHL), where responses were consistently identifiable in both groups. Within-group comparisons between click and CE-Chirp stimulation were performed using the Wilcoxon signed-rank test separately in hearing-impaired and control children. To assess whether the magnitude of the click–chirp difference differed between groups, individual difference scores ($\Delta = \text{click} - \text{CE-Chirp}$) were calculated for latency and amplitude measures, and between-group comparisons were conducted using the Mann–Whitney U test. Wave V amplitude values were log-transformed prior to analysis to reduce skewness and stabilize variance. All analyses of waveform morphology and response features were conducted separately for adult and pediatric cohorts, taking into account differences in age, auditory system maturation, and ABR characteristics.

2.8. Statistical Significance

All statistical tests were two-tailed, and a p -value < 0.05 was considered statistically significant.

3. Results

3.1. Audiometric Results of Adult Group

The overall sample included adult subjects with ski-slope hearing loss and a normal-hearing control cohort, analyzed at the ear level (Table 1).

Table 1. Demographic and audiometric features of the studied cohorts.

Cohort	Group	N (Ears)	Age (Mean $\pm$ SD)	Male/Female	Right/Left	PTA4 (dB HL)
Adults	Ski-slope HL (overall)	21	64.14 $\pm$ 12.39 years	15/6	10/11	31.43 $\pm$ 9.86
Adults	DROP 1 kHz	9	69.56 $\pm$ 10.64 years	5/4	3/6	41.25 $\pm$ 5.04
Adults	DROP 2 kHz	12	60.08 $\pm$ 12.45 years	10/2	7/5	24.06 $\pm$ 4.50
Adults	Normal-hearing controls	25	30.28 $\pm$ 14.98 years	10/15	12/13	6.88 $\pm$ 4.98
Children	Hearing-impaired	7	4.29 $\pm$ 4.79 months	6/1	3/4	Not available
Children	Normal-hearing controls	7	2.00 $\pm$ 0.82 months	3/4	4/3	Not available

Abbreviations: HL, hearing loss; PTA4, pure-tone average across 0.5, 1, 2, and 4 kHz (dB HL); SD, standard deviation.

The adult ski-slope hearing-loss cohort comprised 21 ears, from 15 males and 6 females, with a mean age of 64.1 ± 12.4 years. Laterality distribution included 10 right ears and 11 left ears. Pure-tone average (PTA), calculated across 500, 1000, 2000, and 4000 Hz, was 31.4 ± 9.9 dB HL. The DROP 1K subgroup included nine ears, from five males and four females, with a mean age of 65.9 ± 17.9 years. Laterality distribution included two right ears and seven left ears. Mean PTA4 was 36.1 ± 10.8 dB HL (see Figure 1A). The DROP 2K subgroup comprised 12 ears, from 10 males and 2 females, with a mean age of 62.8 ± 6.4 years. Laterality distribution included eight right ears and four left ears. Mean PTA4 was 24.6 ± 5.4 dB HL (see Figure 1B).

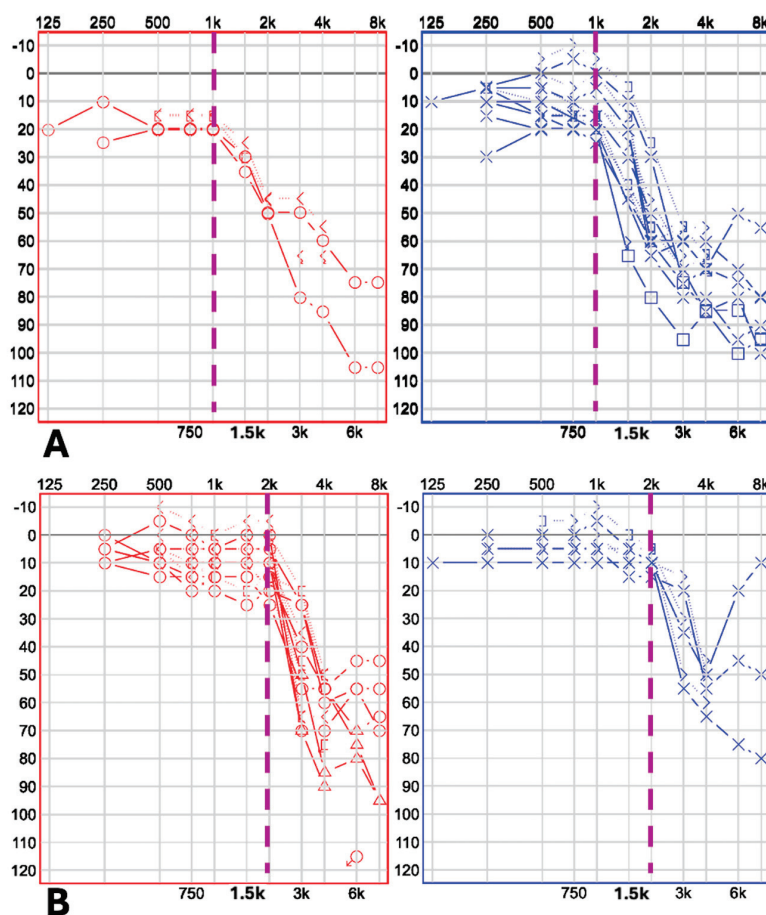


Figure 1. Audiometric threshold configurations of the ears (red for the right ear, blue for the left ear) included in the study, with the panel divided into (A) with all DROP 1 kHz thresholds and (B) with all DROP 2 kHz thresholds.

The normal-hearing control cohort consisted of 25 ears, from 10 males and 15 females, with a mean age of 30.3 ± 15.0 years. Laterality distribution included 12 right ears and 13 left ears. PTA4 values were not reported for this group, as all subjects had hearing thresholds within normal limits and were therefore not directly comparable to the hearing-impaired cohort.

3.2. Audiometric Results of Pediatric Group

The pediatric cohort consisted of 14 children, including 7 hearing-impaired (HI) and 7 normal-hearing (NH) participants (Table 1). The mean age at testing for the entire cohort was 3.1 ± 3.5 months (mean $\pm$ SD). Nine children were male, and five were female. Testing was performed on seven right ears and seven left ears.

In the hearing-impaired group, the mean age was 4.3 ± 4.8 months, with six males and one female; three right ears and four left ears were tested.

In the normal-hearing control group, the mean age was 2.0 ± 0.8 months, including three males and four females; four right ears and three left ears were tested.

3.3. ABR Findings in Adults

ABR threshold estimates obtained with broadband CE-Chirp stimulation were significantly lower than those obtained with click stimulation in the overall hearing-impaired cohort (Wilcoxon signed-rank test, $p = 0.004$). Median ABR threshold decreased from 60 dB nHL with click stimulation to 40 dB nHL with CE-Chirp stimulation (Table 2), with threshold differences occurring in discrete 20 dB steps, reflecting the stimulation protocol. A threshold improvement of at least one 20 dB step was observed in 55.5% of subjects, whereas no difference was found in 38.9%, and a worse threshold with CE-Chirp was observed in a single case. Subgroup analysis revealed that the threshold advantage of CE-Chirp was particularly evident in subjects with hearing loss onset at 2 kHz (DROP 2 kHz group; $p = 0.006$), while no statistically significant difference was observed in subjects with earlier onset of hearing loss (DROP 1 kHz group; $p = 0.293$).

Table 2. Data analysis of the cohorts in terms of wave V comparison in terms of threshold and statistical significance.

Group	N (Ears)	Threshold Click, Median (IQR) (dB nHL)	Threshold CE-Chirp, Median (IQR) (dB nHL)	p	Wave V Present 40 dB (Click), n/N (%)	Wave V Present 40 dB (CE-Chirp), n/N (%)	p (McNemer)
Adults—Ski-slope HL (overall)	21	60 (60–80)	40 (40–60)	<0.001	4/21 (19.0%)	13/21 (61.9%)	0.003
Adults—DROP 1 kHz	9	80 (60–80)	60 (50–80)	0.021	0/9 (0.0%)	2/9 (22.2%)	0.157
Adults—DROP 2 kHz	12	60 (40–60)	40 (40–40)	<0.001	4/12 (33.3%)	11/12 (91.7%)	0.008
Children—Hearing-impaired	7	60 (60–60)	60 (50–60)	0.157	1/7 (14.3%)	2/7 (28.6%)	0.317

Abbreviations: HL, hearing loss; IQR, interquartile range; nHL, normalized hearing level. Notes: ABR threshold comparisons were performed using the Wilcoxon signed-rank test. Wave V detectability at 40 dB nHL was compared using McNemar's test for paired binary outcomes.

Mixed-effects logistic regression modeling of wave V detectability showed no significant effect of stimulus type or recording noise level (RNL) on the probability of identifying wave V at a given intensity, indicating comparable detection probabilities for click and CE-Chirp stimuli. In contrast, audiometric groups significantly influenced wave V detectability.

Data about wave V amplitude are reported in Table 3. Wave V amplitude analysis using linear mixed-effects models demonstrated a significant effect of stimulus intensity, with larger amplitudes observed at higher intensities, while increased RNL was associated

with reduced amplitudes. No significant main effect of stimulus type was observed. Sensitivity analyses restricted to suprathreshold intensities (40–80 dB nHL) and including stimulus-by-intensity interaction terms did not reveal any differential amplitude effects between click and CE-Chirp stimulation.

Table 3. Data analysis of the cohorts in terms of wave V latency, amplitude, and statistical significance.

Group	N (Ears)	Latency 60 dB Click	Latency 60 dB CE-Chirp	<i>p</i>	Latency 80 dB Click	Latency 80 dB CE-Chirp	<i>p</i>	Ln Amp 60 dB Click	Ln Amp 60 dB CE-Chirp	<i>p</i>	Ln Amp 80 dB Click	Ln Amp 80 dB CE-Chirp	<i>p</i>
Adults Ski-slope HL (overall)	21	6.41 ± 0.58	6.05 ± 0.47	<0.001	5.75 ± 0.40	5.52 ± 0.35	<0.001	−1.81 ± 0.99	−1.60 ± 0.58	0.333	−1.35 ± 0.46	−1.36 ± 0.47	0.489
Adults DROP 1 kHz	9	6.58 ± 0.52	6.11 ± 0.52	0.004	5.93 ± 0.37	5.61 ± 0.35	0.008	−2.29 ± 0.38	−1.95 ± 0.64	0.317	−1.33 ± 0.38	−1.45 ± 0.39	0.312
Adults DROP 2 kHz	12	6.29 ± 0.59	6.00 ± 0.44	0.002	5.61 ± 0.38	5.45 ± 0.35	0.002	−1.67 ± 1.08	−1.47 ± 0.54	0.496	−1.36 ± 0.52	−1.29 ± 0.54	0.910
Children Hearing-impaired	7	6.63 ± 0.44	6.29 ± 0.32	0.016	5.92 ± 0.36	5.69 ± 0.34	0.016	−2.00 ± 0.23	−1.59 ± 0.28	0.031	−1.66 ± 0.43	−1.61 ± 0.37	0.312

Abbreviations: HL, hearing loss; Ln, natural logarithm; SD, standard deviation. Notes: Ln amplitude refers to the natural log-transformed wave V amplitude values. Within-group comparisons were performed using the Wilcoxon signed-rank test. Latency and amplitude values are expressed as mean ± SD; latency is measured in milliseconds, while amplitude is in microvolts.

Data about wave V latency are reported in Table 3. Wave V latency analysis at suprathreshold intensities (60 and 80 dB nHL) revealed a robust main effect of stimulus type, with significantly shorter latencies for CE-Chirp compared to click stimulation (mean difference ≈ 1.5 ms, $p < 0.001$). As expected, higher stimulus intensities were associated with shorter latencies. Audiometric group and slope index did not show significant main effects; however, exploratory interaction analyses suggested that stimulus-related latency differences varied with intensity and audiometric configuration.

Correlation analyses between ABR thresholds and behavioral audiometric measures showed that both click- and CE-Chirp-derived thresholds correlated with high-frequency PTA (2–4 kHz), with stronger associations observed for CE-Chirp thresholds, as indicated by both Spearman's rho and Kendall's tau-b coefficients. Additionally, CE-Chirp thresholds were significantly correlated with low- (0.5–1 kHz) and middle-frequency (1–2 kHz) PTA values, whereas click thresholds were not.

3.4. ABR Findings in Children

In hearing-impaired children, ABR thresholds obtained with CE-Chirp stimulation were generally equal to or lower than those obtained with click stimulation. Paired comparisons showed a trend toward lower thresholds with CE-Chirp, although statistical significance was not reached due to the limited sample size.

Wave V detectability at low stimulus intensity (40 dB nHL) was further explored using paired binary analysis. At this intensity level, wave V was observed exclusively with CE-Chirp stimulation in one child, whereas no cases showed wave V exclusively with click stimulation. In the remaining children, wave V was either present or absent with both stimuli. The McNemar test did not reveal a statistically significant difference ($p = 0.317$), although the pattern of discordant responses consistently favored CE-Chirp stimulation (Table 3).

Wave V latency was analyzed at suprathreshold intensities (60 and 80 dB nHL), where responses were consistently identifiable in both hearing-impaired and normal-hearing children. In hearing-impaired children, wave V latency was significantly shorter with CE-Chirp stimulation at both 80 dB nHL (Wilcoxon signed-rank test, $p = 0.016$) and 60 dB nHL ($p = 0.031$). Similarly, in normal-hearing children, CE-Chirp stimulation resulted in significantly shorter wave V latency at 80 dB nHL ($p = 0.008$), while at 60 dB nHL a comparable trend was observed without reaching statistical significance ($p = 0.055$).

Between-group comparisons of click–chirp latency difference scores (Δ latency = click – CE-Chirp) revealed no significant differences between hearing-impaired and control children at either intensity level (80 dB: $p = 0.318$; 60 dB: $p = 1.000$), indicating that the magnitude of the latency reduction associated with CE-Chirp stimulation was comparable across groups.

Wave V amplitude analyses were performed on log-transformed values to account for distributional skewness. In hearing-impaired children, CE-Chirp stimulation produced significantly larger wave V amplitudes than click stimulation at 60 dB nHL (Wilcoxon signed-rank test, $p = 0.016$), whereas no significant difference was observed at 80 dB nHL ($p = 0.172$). In contrast, no significant amplitude differences between CE-Chirp and click stimulation were observed in normal-hearing children at either intensity of 80 dB nHL ($p = 0.498$) or 60 dB nHL ($p = 1.000$).

Between-group comparisons of click–chirp amplitude difference scores did not reveal statistically significant differences at either intensity level (80 dB: $p = 0.181$; 60 dB: $p = 0.073$), although a trend toward larger amplitude differences at 60 dB nHL was observed in the hearing-impaired group.

3.5. Quality Assessment Results of ABR Exams

Across the 50 evaluations (5 traces evaluated by each of 10 clinicians), click-evoked traces were selected as the most suitable for a threshold identification in 45 cases (90%).

4. Discussion

The present study examines the behavior of click and CE-Chirp stimuli in ABR recordings in the specific context of ski-sloping hearing loss, with the aim of improving hearing threshold estimation and informing pediatric diagnostic interpretation. By combining threshold-level and suprathreshold analyses and stratifying subjects according to audiometric configuration, the study provides a framework for understanding how stimulus-dependent ABR features interact with cochlear frequency-specific dysfunction.

Broadband stimuli such as clicks and CE-Chirps do not provide frequency-specific threshold information but rather reflect the functional contribution of the most responsive cochlear regions. The aim of the present study is therefore to compare how these commonly used broadband stimuli behave in the presence of a ski-sloping audiometric configuration.

In adults with ski-sloping hearing loss, CE-Chirp stimulation resulted in lower ABR threshold estimates compared with click stimulation in a substantial proportion of cases, particularly in subjects with preserved hearing up to 2 kHz (DROP 2 kHz group). This finding aligns with the stronger correlations observed between CE-Chirp-derived thresholds and behavioral PTA values across low-, mid-, and high-frequency ranges.

In contrast, click-derived thresholds were only associated with high-frequency PTA. These results suggest that CE-Chirp stimulation may provide a more global representation of cochlear function when mid-frequency regions are still functional. This observation is consistent with the physiological principles underlying chirp stimuli. CE-Chirp was specifically designed to compensate for cochlear travel-time delays by temporally aligning frequency components, thereby enhancing neural synchrony across a broader cochlear region [3,4,6]. Conversely, although click stimuli are broadband, their ABRs predominantly reflect activity originating from the basal cochlea, typically corresponding to the 2–4 kHz region [2].

In the presence of a steep high-frequency hearing loss, this basal bias may lead to ABR threshold estimates that overrepresent high-frequency dysfunction and underestimate residual low- and mid-frequency hearing, as previously described in sloping audiometric configurations [19].

Importantly, the advantage of CE-Chirp stimulation was not observed uniformly across all audiometric profiles. In subjects with hearing loss onset at 1 kHz (DROP 1 kHz group), no significant threshold differences were found between stimuli. This suggests that when cochlear dysfunction extends into lower-frequency regions, the benefits of temporal compensation provided by CE-Chirp may be reduced. This finding supports previous observations that chirp efficacy depends not only on hearing loss severity but also on the frequency range of preserved cochlear function [11]. Such observations are further supported by recent evidence reported by Belet et al. (2025), who compared LS CE-Chirp and click stimuli in subjects with high-frequency hearing loss and similarly observed that chirp-based responses provide advantages that are modulated by the distribution of residual cochlear function rather than by hearing loss severity alone [23]. Their results support the view that stimulus-dependent ABR differences should be interpreted in relation to audiometric configuration, reinforcing the physiological basis and clinical relevance of our observations. This emphasizes the importance of audiometric configuration in ABR interpretation.

At suprathreshold intensities, CE-Chirp stimulation consistently produced shorter wave V latencies than click stimulation (1.5 ms as reported in previous reports), a finding that closely mirrors previous reports in both normal-hearing and hearing-impaired populations [7,24]. This latency reduction reflects enhanced neural synchrony and confirms that the chirp stimulus effectively aligns cochlear responses even in the presence of altered cochlear mechanics. However, no significant amplitude advantage for CE-Chirp stimulation was observed at suprathreshold levels in adults. This pattern is consistent with prior evidence indicating that, at higher intensities, chirp stimuli may induce spread of excitation upward, leading to partial neural desynchronization and attenuation of amplitude benefits [25,26].

This intensity-dependent behavior supports the notion that CE-Chirp stimulation is optimally suited for near-threshold recordings, whereas click stimulation retains advantages in waveform robustness at higher stimulation levels.

The pediatric results, although limited by sample size, followed trends comparable to those observed in adults and were largely consistent with existing pediatric literature. In hearing-impaired children, CE-Chirp-derived thresholds were generally equal to or lower than click-derived thresholds, and wave V latencies were significantly shorter with CE-Chirp stimulation in both hearing-impaired and normal-hearing children. These findings agree with previous studies demonstrating improved neural synchrony, larger amplitudes, and reduced test time with chirp stimuli in neonates and infants [9,27,28].

The observed pediatric latency and amplitude patterns should also be interpreted considering auditory system maturation. ABR wave latencies are known to be prolonged in early infancy, particularly at higher frequencies, and wave amplitudes show age-dependent growth during the first years of life [29,30]. In this developmental context, CE-Chirp stimulation may facilitate wave V identification at lower intensities by recruiting a broader frequency range and optimizing neural synchrony, thereby supporting more reliable threshold estimation when behavioral measures are unavailable. Since behavioral audiometry cannot be used as a reliable comparative benchmark in infants, the adult cohort provides an important interpretative framework for contextualizing the stimulus-dependent ABR patterns observed in the pediatric recordings. However, when considering the relative differences between click and CE-Chirp responses within the same subject, the observed stimulus-dependent patterns cannot be fully explained by maturational factors alone. Given that steep high-frequency losses alter basilar membrane excitation patterns and prolong wave V latency in click-evoked responses, discrepancies between click and CE-

Chirp-derived ABR measures in children should be interpreted with caution and may reflect underlying audiometric configuration rather than maturational effects alone.

Despite these physiological advantages, the clinical usability of the two stimuli differed. In blinded trace evaluations, experienced clinicians overwhelmingly preferred click-evoked waveforms for threshold identification. This finding highlights a critical and often underappreciated aspect of ABR diagnostics: waveform interpretability and examiner confidence remain central to clinical decision-making. In this context, it is important to note that recent advancements aimed at improving acquisition efficiency, such as parallel ABR (pABR), primarily address recording time rather than the underlying physiological interaction between stimulus type and cochlear activation patterns. Therefore, understanding how different stimuli behave in specific audiometric configurations remains essential for accurate interpretation, regardless of recording speed. While CE-Chirp stimulation may offer closer correspondence with behavioral thresholds, click stimulation continues to provide sharper waveform morphology and greater visual clarity, particularly in complex audiometric configurations such as ski-sloping hearing loss.

Despite the value of including a relatively understudied population of patients with ski-sloping hearing loss, this study has several limitations. First, the pediatric cohort was relatively small, reflecting the practical constraints associated with extended ABR testing protocols in early infancy. Consequently, analyses in the pediatric cohort should be regarded as hypothesis-generating rather than confirmatory. A further limitation of the study is that adult-onset ski-sloping hearing loss may differ qualitatively from pediatric-onset hearing loss in terms of underlying pathophysiology. Age-related changes and cumulative environmental exposures likely affect the audiometric configuration in adults, limiting the direct physiological comparability of adult ABR data to pediatric responses. Second, in children, a complete frequency-specific audiological profile is not yet available due to age-related constraints of our cohort (but we chose a cohort that could be realistic for the everyday ABR clinical use). Third, a subset of adult cases could not be included in the statistical analyses because of missing data (thus limiting the cohort of adults). In addition, stratification of the adult hearing-impaired cohort into DROP 1 kHz and DROP 2 kHz subgroups resulted in relatively small subgroup sizes. Subgroup-related findings should therefore be interpreted cautiously and considered exploratory. Fourth, only CE-Chirp stimuli were evaluated, and LS-Chirp stimuli were not tested (and this might have revealed even additional data). Fifth, ABR threshold estimation was performed using 20 dB step sizes rather than finer intensity increments (e.g., 5–10 dB), which may have reduced threshold resolution. Therefore, the results should be interpreted in terms of relative differences between stimuli rather than precise threshold estimation. Finally, our protocol did not include the use of frequency-specific stimuli such as tone pips or tone bursts. While broadband chirps are highly effective for maximizing neural synchrony and identifying a global electrophysiological threshold, the absence of frequency-specific testing limits our ability to provide a detailed mapping of the audiometric configuration across discrete frequencies. In cases of sloping hearing loss, the ABR threshold obtained with broadband stimuli may primarily reflect the hearing sensitivity of the most preserved cochlear regions, potentially leading to an underestimation of hearing loss at poorer-frequency regions.

Moreover, the inclusion of both adult and pediatric cohorts introduces inherent methodological constraints. Adult ABR recordings were used as a reference model in a well-defined audiometric condition, in the absence of comparable behavioral thresholds in the pediatric cohort, but they cannot be considered a direct physiological proxy for pediatric responses due to known differences in auditory system maturation and recording optimization strategies. Therefore, cross-population interpretations should be made with caution.

From a clinical standpoint, discrepancies between click- and CE-Chirp-derived ABR thresholds in children should therefore not be interpreted solely as maturational effects or differences in neural synchrony. Rather, such discrepancies may represent a potential indicator of an underlying ski-sloping hearing loss. In the absence of alternative clinical explanations, these findings should prompt further frequency-specific evaluation, including tone-burst ABR or auditory steady-state response (ASSR), with particular attention to the mid-to-high frequency range.

Overall, the present study supports a complementary use of click and CE-Chirp stimuli in ABR assessment. Click stimulation remains indispensable for reliable waveform identification and clinical confidence (i.e., otoneurologic evaluation of the neural auditory pathway), whereas CE-Chirp stimulation provides advantages in threshold estimation and neural synchrony, particularly near threshold (i.e., in ski-slope patients) and in pediatric settings (i.e., in preterm infants for improved threshold definition). An integrated, stimulus-specific approach may therefore improve diagnostic accuracy and support more informed rehabilitative planning in children with suspected high-frequency hearing loss. The discrepancy between click- and CE-Chirp-derived ABR thresholds may also represent a potentially useful clinical cue for non-flat audiometric configurations, warranting further investigation with frequency-specific measures.

5. Conclusions

This study highlights the complementary value of click and CE-Chirp stimuli in ABR-based hearing threshold estimation, especially for ski-sloping hearing loss.

Click stimulation remains a robust and reliable reference, providing clear and highly interpretable waveforms across a wide range of intensities, even in complex audiometric profiles. CE-Chirp stimulation, by enhancing neural synchrony and engaging broader cochlear regions, may provide threshold estimates closer to overall hearing sensitivity. This is particularly relevant near threshold and in pediatric assessments, where low-intensity stimuli help minimize discomfort and ensure reliability in auditory threshold determination.

However, acknowledging the sample size and the complexity of cochlear hearing loss configurations, these results should be interpreted as a validation of the clinical methodology and a hypothesis-generating framework rather than definitive evidence of stimulus specificity.

These findings support the integration of both stimuli into ABR protocols. A combined approach may improve diagnostic accuracy across diverse patient populations, addressing the specific needs of both adult and pediatric assessments, especially for high-frequency hearing loss. Future studies involving larger pediatric cohorts and frequency-specific outcome measures are warranted to further refine ABR protocols and optimize clinical decision-making. In addition, future research should explore the potential advantages of Level-Specific (LS) CE-Chirp stimuli, which adapt temporal compensation according to stimulation level. Comparative investigations between standard and LS-Chirp paradigms may therefore help clarify the role of stimulus design in configuration-dependent ABR interpretation.

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Abbreviations

The following abbreviations are used in this manuscript:

ABR	Auditory Brainstem Response
IMT	Italian Matrix Test
DPOAE	Distortion Product Otoacoustic Emissions
RNL	Recording Noise Level
NHL	Normalized Hearing Level
NH	Normal Hearing
SD	Standard Deviation
HI	Hearing Impaired
PTA	Pure Tone Average

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Article

Unilateral Sensorineural Hearing Loss in Children: Etiology, Audiological Characteristics, and Treatment

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Abstract: The aim of this study was to evaluate audiological characteristics and parents' opinions on hearing device use in children with unilateral sensorineural hearing loss (USNHL) who attended a tertiary-level audiologic center. The medical charts of 70 children aged 6 to 12 years with USNHL were reviewed. In 51.4% of cases, the children were diagnosed with USNHL after the age of 2 years. The main causes of USNHL were congenital cytomegalovirus infection (21.4%) and unilateral cochlear nerve hypoplasia (12.9%). The percentage of patients wearing a hearing device was 45.7% (32/70); of these, 28 (87.5%) wore a conventional hearing aid, 2 (6.3%) a CROS device, and 2 (6.3%) a cochlear implant. Regarding the choice to use a hearing device, no significant differences were found between the subcategories of hearing loss degree ($p = 0.55$) and audiometric configuration ($p = 0.54$). Most parents of children with mild-to-severe USNHL observed improved attention (90.9%), and reduced fatigue and restlessness (86.4%) using the hearing aid. These children performed significantly better on all audiological tests (speech perception in quiet and in noise conditions, and sound localization) while wearing the hearing aid ($p < 0.001$). More efforts should be made to raise awareness among professionals and parents about the negative consequences of uncorrected USNHL.

Keywords: unilateral hearing loss; hearing aid; cochlear implant; children

1. Introduction

Unilateral hearing loss (UHL) is defined as normal hearing in one ear and impaired hearing in the contralateral ear. According to the “National Workshop on mild and unilateral hearing loss”, permanent UHL is characterized by an average pure tone air conduction threshold at frequencies of 500 Hz, 1000 Hz, and 2000 Hz ≥ 20 decibels Hearing Level (dB HL) or pure tone air conduction thresholds > 25 dB HL at two or more frequencies above 2000 Hz in the impaired ear; meanwhile, the average pure tone air conduction threshold in the unaffected ear should be ≤ 15 dB [1]. Hearing loss (HL) can range from mild to profound, and have different audiometric configurations (flat, rising, or sloping) [2]. Specifically, single-sided deafness (SSD) refers to profound sensorineural HL or nonfunctional hearing in one ear, while the other ear maintains normal hearing [3]. The prevalence of congenital UHL is about 0.3–1 per 1000 newborns, with a remarkable increase (1.2–4.6 per 100) in newborns admitted to the neonatal intensive care unit [4]. However, it is important to underline that most children with mild HL and those with late-onset or acquired HL are not identified by universal newborn hearing screening [1,5]. Moreover, a recent study by Fitzpatrick et al. reported that nearly half of children with UHL are at risk of experiencing further hearing decline in the affected ear or developing bilateral HL [6]. The prevalence of

UHL increases with age; it is estimated that 7.2% of adults in the United States of America are affected by UHL [7]. The most common causes of unilateral sensorineural hearing loss (USNHL) include premature birth, congenital or postnatal infections (especially congenital cytomegalovirus and meningitis), structural anomalies (such as cochlear nerve hypoplasia/aplasia and enlarged vestibular aqueduct), genetic underpinnings, and temporal bone trauma [6,8–12]; however, in many cases, the etiology remains unknown [9]. A review by Lieu showed that UHL in children has a negative impact on speech–language development, cognition, and quality of life [13]. Furthermore, Bess et al. demonstrated that children with UHL are more susceptible to listening-related fatigue and that their fatigue is akin to that of pediatric patients with bilateral HL [14]. In the past, it was traditionally believed that unilateral hearing was the minimum requirement for acceptable speech development and that, consequently, the use of hearing aids was not necessary [15]. However, since the 1980s, significant difficulties have been reported for children with UHL in the areas of localization, listening in noise, language, and academic functions [16]. In particular, Schmithorst et al. suggested that monaural hearing influences the development of brain networks associated with cross-modal sensory processing and the regulation of the default network during spoken language processing [17]. Currently, published evidence and clinical experience confirm the crucial role of the timely diagnosis and treatment of UHL in improving linguistic, cognitive, socio-emotional, and communicative development [15]. Although the treatment of USNHL is still controversial, recent comprehensive reviews have suggested that, in selected cases, cochlear implantation for children with SSD can lead to significant improvements in overall hearing outcomes [18,19]. The aim of this study was to evaluate the audiological characteristics (degree of HL, affected side, audiometric configuration, and hearing outcomes), adherence to treatment (hearing aid or cochlear implant), causes of HL, and parents' opinions on hearing device use in children aged 6 to 12 years with USNHL who attended our tertiary-level referral audiologic center.

2. Materials and Methods

2.1. Data Collection

In the present study, all medical reports and charts of children aged 6 to 12 years with USNHL who were referred to the tertiary-level audiology center of the Fondazione IRCCS Ca' Granda, Ospedale Maggiore Policlinico (Milan, Italy) in the period from 1 January 2021 to 31 December 2023 were reviewed.

Specifically, the study included all children who met the following criteria:

- Children 6 through 12 years of age with a confirmed diagnosis of USNHL;
- Children with at least 1 year of hearing aid or cochlear implant use in the impaired ear, or children who had never received any hearing treatment;
- Children who had undergone a comprehensive audiological evaluation, including otomicroscopy, tympanometry, reflex threshold measurements, pure tone audiometry (covering frequencies from 250 Hz to 8000 Hz), speech audiometry, Simplified Italian Matrix Test (SiIMax), and Sound Localization Test;
- Children who had undergone video head impulse test (v-HIT), high-resolution magnetic resonance imaging (MRI) of the brain, high-resolution cone beam computed tomography (CBCT) of the head, and genetic testing.

Specifically, genetic testing was performed by gene amplification (polymerase chain reaction) of the entire coding region of the 101 genes investigated, followed by sequencing analysis using next-generation sequencing (NGS) technology. The list of genes investigated was as follows: ACTG1, AIFM1, ATP6V1B1, BSND, CCDC50, CDH23, CEACAM16, CIB2, CISD2, CLDN14, CLRN1, COCH, COL11A1, COL11A2, COL2A1, COL9A1, COL9A2, CRYM, DCDC2, DFNAS (GSDME), DFNB31 (WHRN), DFNB59, DIABLO, DIAPH1, DIAPH3, EDN3, EDNRB, ESPN, ESRRB, EYA1, EYA4, FOXI1, GIPC3, GJB2, GJB3, GJB6, GPR98 (ADGRV1), GRHL2, GRXCR1, HARS, HGF, HOMER2, ILDR1, KARS, KCNE1, KCNJ10, KCNQ1, KCNQ4, KITLG, LHFPL5, LOXHD1, LRTOMT, MARVELD2, MET, MIR96, MITE, MSRB3, MYH14, MYH9, MYO15A, MYO3A, MYO6, MYO7A, NARS2, OPA1,

OTOA, OTOF, PAX3, PCDH15, PDZD7, POLR1C, POLR1D, POU3F4, POU4F3, PRPS1, PTPRQ, RDX, SERPINB6, SIX1, SIX5, SLC17A8, SLC26A4 (PDS), SLC26A5, SMPX, SNAI2, SOX10, STRC, TBC1D24, TCOF1, TECTA, TJP2, TMC1, TMIE, TMPRSS3, TNC, TPRN, TRIOBP, USH1C, USH1G, USH2A, and WFS1.

Although this genetic test is very accurate, some limitations of the diagnostic method must be considered, including the inability to detect (1) other genetic diseases or genes not specifically investigated; (2) mutations located in intronic regions beyond ± 5 nucleotides from breakpoints; and (3) deletions, inversions, or duplications greater than 20 base pairs.

Family history regarding genetic predisposition to hearing impairment was also collected. The study excluded all children with the following:

- Conductive or mixed HL;
- Congenital malformations or acquired pathologies of the outer or middle ear;
- Genetic syndromes;
- Any type of bilateral HL;
- Cognitive delay.

Degrees of HL were classified according to pure-tone averages at 500 Hz, 1000 Hz, 2000 Hz, and 4000 Hz (PTA-4). The categories of HL were distinguished into the following: mild HL, which ranged from 26 to 40 dB HL; moderate HL, from 41 to 60 dB HL; severe HL, from 61 to 80 dB HL; and profound HL, greater than 80 dB HL [20]. Regarding USNHL, the following variables were specifically investigated: age at diagnosis, affected side, HL degree, hearing changes after diagnosis, audiometric configuration, and use of a hearing device (conventional behind-the-ear [BTE] hearing aid, wireless contralateral-routing-of-signal [CROS] device, or cochlear implant). The causes of each child's USNHL and possible associations between hearing device use and gender, degree of HL, or type of audiometric configuration were also evaluated. Additional variables investigated in children using a hearing device were as follows: age at first hearing device fitting, time elapsed between HL diagnosis and hearing device fitting, and mean daily hours of hearing device use (by analyzing the data logging of the device software). Moreover, we compared audiological test scores (PTA-4, intellection threshold, SiIMax, and Sound Localization Test) obtained without and with the hearing aid by children with mild-to-severe USNHL who regularly used a hearing aid. The "intellection threshold" was defined as the level at which the subject understood 100% of the words [21].

- *Simplified Italian Matrix Test (SiIMax)*

The SiIMax speech material included a subset of items (7 numerals, 7 nouns, and 7 adjectives) from the original 50-word base matrix of the ITAMatrix test, which means that shorter speech phrases, such as "seven white balls", were used rather than full sentences. Specifically, each test list consisted of 14 randomized three-word sentences. Two loudspeakers were positioned at head height, respectively, 1 m in front of and behind the child sitting inside the sound booth; background noise was set at 65 dB pressure level and presented at 0° and 180° azimuth, while speech was presented at 0° azimuth and automatically adjusted according to the child's previous response. The result of the test was reported as a score representing the signal-to-noise ratio (SNR) in dB at which the child could recognize 50% of the speech material (speech recognition threshold, SRT) [22,23].

- *Sound Localization Test*

Sound localization was tested by means of 8 loudspeakers arranged in a full circle with an angular distance of 45°; all loudspeakers were positioned at head height and labeled from number 1 to number 8. Participants were placed centrally inside the sound booth, comfortably seated on a chair, facing the loudspeaker at 0° azimuth (loudspeaker number 1), and equidistant, 1 m away, from the different loudspeakers [24]. Children were instructed not to move their head during the presentation of the auditory stimulus (a 1000 Hz warble tone presented at 40 dB sensation level for a duration of 3 s); immediately after the end of each single auditory stimulus, they had to indicate the perceived sound source by vocally reporting the number of the loudspeaker (for this purpose, they were

temporarily allowed to turn their head to read the number of the loudspeaker). The level of auditory stimuli was kept fixed, as variations in intensity appeared to have no impact on sound localization [25]. The result of the test was reported as the percentage of correct responses, that is, the number of times a participant accurately identified the loudspeaker presenting the sound ($n = 0-8$) divided by the total number of presentations ($n = 8$, one per loudspeaker), multiplied by 100.

Both the SiMax and the Sound Localization Test were performed only once after a brief trial, as these audiological examinations were explained in detail to the children by experienced audiologists before being performed.

- *Parents' opinions on children's use of hearing devices*

Parents of hearing-aid-wearing children with mild-to-severe USNHL were asked for their opinions on the impact of regular hearing aid use on their child's selective attention (*"How do you perceive your child's selective attention when he/she constantly wears the hearing aid during the day?"*), fatigue (*"How do you perceive your child's fatigue when he/she constantly wears the hearing aid during the day?"*), restlessness (*"How do you perceive your child's restlessness when he/she constantly wears the hearing aid during the day?"*), and overall hearing ability (*"How do you perceive your child's hearing ability when he/she constantly wears the hearing aid during the day?"*) compared to when their child did not use the hearing aid (i.e., before hearing aid fitting, during sports activities or other special circumstances, or because the hearing aid was out of power or broken). Each question had five possible responses on a five-item Likert scale: greatly worsened = 1, worsened = 2, same = 3, improved = 4, and greatly improved = 5.

The following definitions were used [26–28]:

- Selective attention: child's ability to focus on important environmental stimuli while simultaneously suppressing irrelevant or distracting information;
- Fatigue: child's sense of tiredness, lack of energy, and feeling of exhaustion;
- Restlessness: child's feeling of irritability and agitation;
- Hearing abilities: parental perception of the child's hearing.

The same questions were asked to parents of children with profound USNHL who regularly used a hearing device. Children's personal opinion of their overall hearing ability when wearing a hearing device was also analyzed (using the five-item Likert scale previously described). Finally, parents of children with mild-to-severe USNHL who did not use any hearing aids were asked the reasons for that choice, while parents of children with profound USNHL were asked why their child had never undergone cochlear implantation until then. This retrospective study was conducted in accordance with the World Medical Association Declaration of Helsinki, and received approval from the local ethics committee and parental consent.

2.2. Statistical Analysis

Stata 17 software (StataCorp., College Station, TX, USA, 2021) was used to perform the statistical analyses. Categorical variables were analyzed using the chi-squared test. The difference between audiological test scores without and with the hearing aid was assessed using the nonparametric Wilcoxon signed-rank sum test. A p -value of <0.05 was considered statistically significant.

3. Results

A total of 70 children with USNHL, including 37 (52.9%) males and 33 (47.1%) females, met the inclusion criteria. The mean age of the study population was 8.8 ± 2.5 years, and the PTA-4 of the ear affected by USNHL was 71.1 ± 25.1 dB HL. Table 1 provides a summary of the main characteristics of USNHL observed in the study population. Among children with USNHL, 23 (32.9%) had unilaterally failed the universal neonatal hearing screening. The main causes of USNHL in the study population were congenital cytomegalovirus infection ($n = 15$, 21.4%) and unilateral cochlear nerve hypoplasia ($n = 9$, 12.9%), while,

in 40% of patients ($n = 28$), the etiology remained unknown (Table 2). No child in the study had complete cochlear nerve aplasia, or a family history of genetic predisposition to HL. Unilateral vestibular hypofunction, detected by bedside vestibular examination and confirmed by v-HIT, was found in 14 (20%) patients; of these, 6 had profound USNHL, 5 severe USNHL, 2 moderate USNHL, and 1 mild USNHL. No significant correlation was found between the presence of unilateral vestibular hypofunction and degree of USNHL ($p = 0.73$). Regarding the choice to use a hearing device, no significant differences were found between males and females ($p = 0.6$) or between the subcategories of HL degree ($p = 0.55$) and audiometric configuration ($p = 0.54$) (Table 3). Table 4 shows the temporal characteristics of hearing treatment in children with mild-to-severe USNHL (Table 4). The mean time elapsed between HL diagnosis and hearing aid fitting was 22.0 ± 15.5 months. The mean daily hours of hearing aid use were 10.3 ± 2.8 . Children with mild-to-severe USNHL who regularly used a hearing aid performed significantly better on all audiological assessments (pure tone audiometry, speech audiometry, SiMax, and Sound Localization Test) while wearing the hearing aid ($p < 0.001$) (Table 5). Parents observed a positive impact of the hearing aid on the child's selective attention, fatigue, restlessness, and overall hearing ability in 90.9%, 86.4%, 86.4%, and 90.9% of cases, respectively (Table 6). Among these children with mild-to-severe USNHL, 7 (31.8%) and 15 (68.2%) reported better and much better hearing with regular use of the hearing aid, respectively. The main reasons for not using a hearing aid among children with mild-to-severe USNHL were aesthetic issues ($n = 9$, 34.6%), followed by the fact that this solution had never been proposed before ($n = 6$, 23.1%) (Figure 1). Among children with profound USNHL, 2 (9.1%) had undergone cochlear implantation, 6 (27.3%) were fitted with a conventional BTE hearing aid, and 2 (9.1%) were fitted with a CROS hearing aid, while 12 (54.5%) received no hearing treatment. None of the patients included in the study used non-surgical or surgical bone conduction devices. The reasons why 90.1% (20/22) of children with profound USNHL did not undergo cochlear implantation were the following: this solution had never been proposed before ($n = 10$, 50%), aesthetic issues ($n = 5$, 25%), and parents' concerns about surgery ($n = 5$, 25%). Table 7 shows the temporal characteristics of hearing treatment, the results of audiological tests without and with the hearing device, and parents' opinions of children with profound USNHL who regularly used a hearing device (Table 7). Subjectively, both patients with a cochlear implant reported much better hearing (rated 5 out of 5 points) with regular use of the device.

Table 1. Characteristics of unilateral sensorineural hearing loss in the study population.

Variables	Patients, N (%)
Age at diagnosis	
<6 months	25 (35.7)
6–24 months	9 (12.9)
25–60 months	20 (28.6)
>60 months	16 (22.9)
Side	
Right	37 (52.9)
Left	33 (47.1)
Degree	
Mild	8 (11.4)
Moderate	14 (20.0)
Severe	26 (37.1)
Profound	22 (31.4)

Table 1. Cont.

Variables	Patients, N (%)
Hearing changes after diagnosis	
No changes	58 (82.9)
Improvement	2 (2.9)
Deterioration	10 (14.3)
Audiometric configuration	
Rising	6 (8.6)
Flat	40 (57.1)
Sloping	24 (34.3)
Hearing device	
None	38 (54.3)
Hearing aid	30 (42.9)
Cochlear implant	2 (2.9)
TOTAL	70 (100.0)

Table 2. Causes of unilateral sensorineural hearing loss in the study population.

Etiology	Patients, N (%)
(1) Infectious	21 (30.0)
Congenital cytomegalovirus	15 (21.4)
Epstein–Barr virus	4 (5.7)
Bacterial meningitis	2 (2.9)
(2) Otologic	15 (21.4)
Cochlear nerve hypoplasia	9 (12.9)
Enlarged vestibular aqueduct	4 (5.7)
Superior semicircular canal dehiscence	2 (2.9)
(3) Other causes	6 (8.6)
Prematurity	4 (5.7)
Pathogenic mutations in GJB2 gene	2 (2.9)
(4) Unknown	28 (40.0)
TOTAL	70 (100.0)

Table 3. Demographic and audiological characteristics of patients with unilateral sensorineural hearing loss using a hearing device.

Variables	Total (N)	Patients with a Hearing Device, N (%)	<i>p</i> -Value
Gender			
Male	37	18 (48.6)	0.6
Female	33	14 (42.4)	
Degree of hearing loss			
Mild	8	2 (25.0)	0.55
Moderate	14	6 (42.9)	
Severe	26	14 (53.8)	

Table 3. Cont.

Variables	Total (N)	Patients with a Hearing Device, N (%)	<i>p</i> -Value
Profound	22	10 (45.5)	
Audiometric configuration			
Rising	6	2 (33.3)	0.54
Flat	40	17 (42.5)	
Sloping	24	13 (54.2)	
TOTAL	70	32 (45.7)	

Table 4. Temporal characteristics of hearing treatment in children with mild-to-severe unilateral sensorineural hearing loss.

Variables	Patients, N (%)
Age at first hearing aid fitting	
<6 months	0 (0.0)
6–24 months	2 (9.1)
25–60 months	10 (45.5)
>60 months	10 (45.5)
Time elapsed between hearing loss diagnosis and hearing aid fitting	
<6 months	2 (9.1)
6–12 months	6 (27.3)
13–36 months	11 (50.0)
>36 months	3 (13.6)
Daily hours of hearing aid use	
<6 h	1 (4.5)
7–10 h	9 (40.9)
>10 h	12 (54.5)
TOTAL	22 (100.0)

Table 5. Hearing tests performed without and with the hearing aid by children with mild-to-severe unilateral sensorineural hearing loss who regularly used a hearing aid.

	Without Hearing Aid (Mean $\pm$ sd)	With Hearing Aid (Mean $\pm$ sd)	<i>p</i> -Value
PTA-4 (dB)	60.4 $\pm$ 10.1	30.2 $\pm$ 6.8	<0.001
Intellection threshold (dB)	80.5 $\pm$ 16.2	40.5 $\pm$ 7.9	<0.001
Simplified Italian Matrix Test (dB)	−1.8 $\pm$ 1.0	−4.3 $\pm$ 0.9	<0.001
Sound Localization Test (%)	51.7 $\pm$ 15.1	90.9 $\pm$ 8.8	<0.001

PTA = pure-tone averages. Intellection threshold = level at which the subject understands 100% of the words.

Table 6. Parents' opinions on the impact of the hearing aid on selective attention, fatigue, restlessness, and overall hearing ability of their children with mild-to-severe unilateral sensorineural hearing loss.

Variables	Parents' Opinions, N (%)
Selective attention [1–5 points]	
Greatly worsened [1]	0 (0.0)
Worsened [2]	0 (0.0)

Table 6. Cont.

Variables	Parents' Opinions, N (%)
Same [3]	2 (9.1)
Improved [4]	7 (31.8)
Greatly improved [5]	13 (59.1)
Fatigue [1–5]	
Greatly worsened [1]	0 (0.0)
Worsened [2]	0 (0.0)
Same [3]	3 (13.6)
Improved [4]	12 (54.5)
Greatly improved [5]	7 (31.8)
Restlessness [1–5]	
Greatly worsened [1]	0 (0.0)
Worsened [2]	0 (0.0)
Same [3]	3 (13.6)
Improved [4]	10 (45.5)
Greatly improved [5]	9 (40.9)
Overall hearing ability [1–5]	
Greatly worsened [1]	0 (0.0)
Worsened [2]	0 (0.0)
Same [3]	2 (9.1)
Improved [4]	8 (36.4)
Greatly improved [5]	12 (54.5)
TOTAL	22 (100.0)

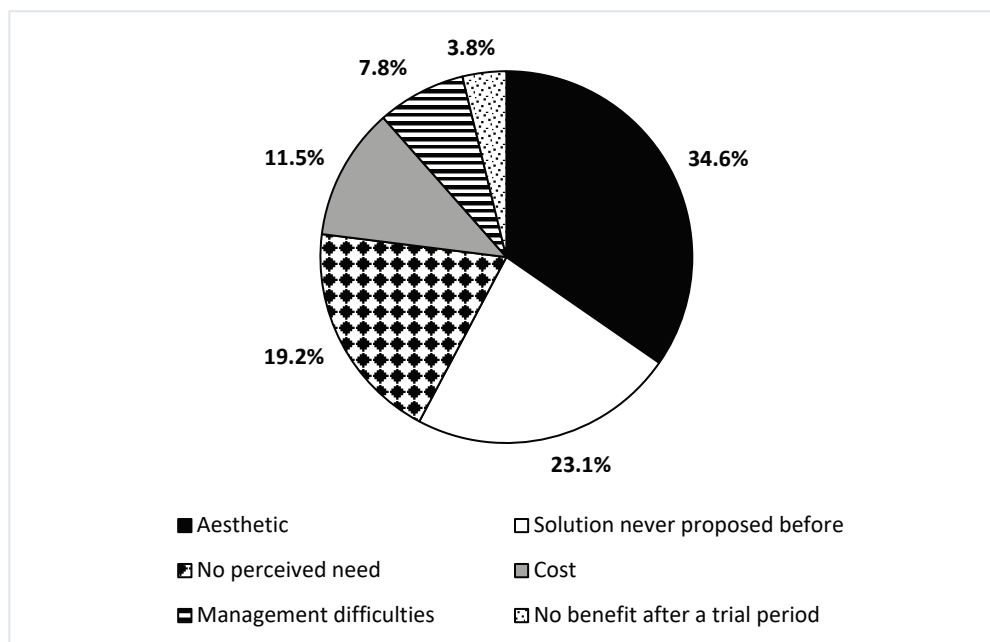


Figure 1. Reasons why children with mild-to-severe unilateral sensorineural hearing loss do not use a hearing aid.

Table 7. Temporal characteristics of hearing treatment, audiological test results, and parents' opinions of children with profound unilateral sensorineural hearing loss using a hearing device.

Variables	Cochlear Implant			Conventional Hearing Aid				CROS		
	P1	P2	P3	P4	P5	P6	P7	P8	P9	P10
Gender	F	M	F	F	M	M	M	M	F	M
Age (years)	7	6	10	8	11	11	11	6	7	8
Side	R	L	L	R	R	R	L	R	L	R
Age at first hearing device fitting (months)	60	48	84	60	60	84	70	36	60	72
Time elapsed between hearing loss diagnosis and hearing device fitting (months)	57	45	20	54	52	14	30	33	57	69
Daily hours of hearing device use (hours)	14	12	10	12	10	10	12	14	8	8
PTA-4 without hearing device (dB)	102.5	107.5	95.5	90	100	96.3	95	88.8	110	105
PTA-4 with hearing device (dB)	25	20	67.5	65	75	70	65	63.8	/	/
Maximum speech intelligibility without hearing device (%)	0	0	20	20	0	10	20	30	0	0
Maximum speech intelligibility with hearing device (%)	100	100	50	40	30	40	50	50	/	/
Simplified Italian matrix test without hearing device (dB)	−1.2	−0.8	−2.4	−1.8	−2.2	−3	−2.2	−1.8	−1.2	−1
Simplified Italian matrix test with hearing device (dB)	−5.2	−5	−3.4	−3.0	−3.2	−3.8	−3.6	−3.2	−2.0	−1.8
Sound Localization Test without hearing device (%)	50	37.5	50	50	37.5	37.5	37.5	50	37.5	37.5
Sound Localization Test with hearing device (%)	100	100	62.5	62.5	62.5	62.5	62.5	87.5	37.5	37.5
Parents' opinions [1–5]										
Selective attention [1–5]	5	5	4	4	4	3	4	4	3	4
Fatigue [1–5]	5	5	4	3	4	4	3	4	4	4
Restlessness [1–5]	5	4	4	4	3	4	4	4	3	3
Overall hearing ability [1–5]	5	5	4	4	3	4	3	4	3	3

F = female. L = left. M = male. P = patient. PTA = pure-tone average. R = right.

4. Discussion

The present study investigated the causes of HL, audiological characteristics, adherence to treatment, and parents' opinions on hearing device use in children aged 6 to 12 years with USNHL who attended a tertiary-level referral audiologic center. Interestingly, more than two-thirds of the study population had bilaterally passed the universal newborn hearing screening, and more than half of children were diagnosed with USNHL after the age of 2 years. These findings could be explained by the high proportion of cases of USNHL due to congenital cytomegalovirus infection, acquired etiologies, and other conditions associated with late-onset or progressive HL. Although universal newborn screening has some limitations [1,5], it should be emphasized that, before its introduction, the percentage of children with USNHL detected before 6 months of age was only 3% [29], compared to 35.7% in our study. In two out of five children, the etiology of USNHL remained unknown, which is consistent with what has been reported by other studies in the literature [29,30] and makes it difficult to predict the long-term hearing outcomes of these patients. Possible undetected causes of USNHL in the pediatric population might be attributable to genetic variants of

unknown/uncertain significance, undiagnosed congenital cytomegalovirus infection (as most cases are asymptomatic at birth), or asymptomatic/paucisymptomatic postnatal infections [5,10,12]. This study showed that the characteristics of UNSHL are highly variable regarding age at diagnosis, affected side, degree, hearing changes over time, audiometric configuration, and use of a hearing device. Moreover, one in five children was diagnosed with unilateral vestibular hypofunction, reinforcing the need for routine vestibular and balance assessment in all pediatric patients with USNHL, as already suggested by previous studies [31,32]. Although several studies have demonstrated the negative consequences and long-term implications of uncorrected USNHL [9,13–16], less than half of our patients used a hearing device. The high percentage of non-users of hearing devices was largely attributed not only to aesthetic concerns due to social stigma, but also to parents' lack of knowledge since this solution had never been proposed before by professionals. Although these reasons may seem surprising, they have also been reported by other studies in the literature [9,33–36], thus suggesting the need for shared decision-making processes, psychosocial supports, public health awareness campaigns, and standardized guidance from audiology services on how to manage pediatric USNHL. In our study, uncertainty about the application and acceptance of a hearing device in children with USNHL also emerged from the long time that often elapses between diagnosis and hearing device fitting, unlike what usually happens for children with bilateral SNHL [36]. In this regard, a population-based study by Fitzpatrick et al. demonstrated that more than half of children with USNHL or mild bilateral SNHL experience a considerable delay from identification to hearing aid fitting, but the reasons for this delay are often unknown [37]. As a matter of fact, parents often perceive professionals' hesitation and lack of conviction about the potential benefits of hearing aids in children with USNHL, as well as professionals' tendency to minimize the importance of milder HL [38]. However, only one of our patients did not use a hearing aid due to lack of benefit after an adequate trial period, confirming that most children with UNSHL who have tried a hearing device continue to use it [9,33]. The subjective benefit of wearing a hearing aid was also indirectly proven by the high mean daily hearing aid use, with all but one of our patients using the device for at least 7 h per day. Interestingly, the choice to wear a hearing device was not influenced by the degree of HL or the type of audiometric configuration. Among patients with mild-to-severe USNHL, the advantages of hearing aid use were demonstrated not only by significant improvements in sound and speech perception in quiet, speech perception in noise, and sound localization, but also by positive feedback from children and parents. In particular, most parents noticed that the use of a hearing aid improved selective attention and reduced fatigue and restlessness in their children. These findings are not surprising since children with USNHL are known to have higher levels of fatigue and irritability, and a lower performance on selective attention tests [39–42].

One of the greatest challenges is counseling families of children with profound USNHL due to the lack of robust clinical data to steer shared decision-making and identify potential benefits of unilateral cochlear implantation [18]. However, parents should be advised that children with SSD often have difficulty with sound localization and speech perception in noisy environments due to the absence of binaural auditory input [43]; in this context, unilateral cochlear implantation should be considered as a possible viable treatment aimed at restoring binaural hearing [44]. Indeed, on 19 July 2019, the Food and Drug Administration (FDA) granted approval for the MED-EL cochlear implant in children aged ≥ 5 years with SSD (maximum auditory deprivation = 10 years); Cochlear Americas received the first approval on 11 January 2022 [43]. The existing FDA guidelines for unilateral cochlear implantation stipulate that the MED-EL device requires a PTA-4 equal to or greater than 90 dB HL, while the Cochlear device requires a PTA-4 greater than 80 dB HL [43].

A recent clinical trial by Brown et al. found that pediatric patients implanted for USNHL achieved marked improvements in their ability to perceive speech in both quiet and noisy environments and to localize sounds; specifically, these children already showed head shadow and summation effects with their cochlear implant as early as 6 months

after activation, whereas, by 12 months after activation, they developed a significant squelch benefit [45]. Although the age limit for cochlear implantation was set at 5 years because of the lack of rigorous clinical studies in younger children, auditory neuroplasticity declines dramatically after the age of 7 years, meaning that potential benefits for individuals with congenital SSD are confined within a short time window [46]. Therefore, it can be reasonably expected that cochlear implantation at a younger age would be beneficial in pediatric patients with profound USNHL. A recent study by Patro et al. demonstrated that children with SSD can safely undergo surgery even under the age of 5, resulting in greater improvements in speech recognition [47]. It is important to underline that some clinical conditions deserve special consideration, regardless of the patient's age. For example, individuals with SSD due to bacterial meningitis should be implanted as soon as possible because swift ossification may obliterate the inner ear and hinder the success of the procedure [43]. Moreover, pediatric patients with SSD at risk of hearing deterioration in the contralateral ear, such as those affected by a congenital cytomegalovirus infection or an enlarged vestibular aqueduct, should promptly receive a cochlear implant not only to restore bilateral auditory input, but also to provide a lifeline in case of sudden worsening of the hearing threshold in the normal ear [5,48]. However, it should be kept in mind that candidacy for unilateral cochlear implantation requires the careful assessment of imaging, audiological testing, duration of SNHL, age, comorbid conditions, psychological support, and parental motivation [18,43]. Alternative hearing technologies available for children with SSD include re-routing devices such as CROS and bone conduction devices, but these solutions cannot provide binaural input, which means they do not enhance sound localization [9,49]. Although these devices help attenuate the head shadow effect, they can also impair speech perception when the noise comes from the impaired ear's side [9]. In particular, CROS hearing aids may have a negative effect in complex listening scenarios if patients are unable to manage the device according to the context, which is why these types of devices are generally not recommended under the age of 5 [50]. Furthermore, it is important to remember that hearing deprivation due to SSD leads to irreversible changes in the auditory cortex, which re-routing devices are unable to prevent because they do not restore hearing in the impaired ear [9]. Given the limited amplification capacity, a conventional BTE hearing aid has few long-term benefits for children with profound USNHL. However, the auditory stimulation of the impaired ear through a well-fitted hearing aid could be useful while waiting for parents to decide on whether to have their children implanted [51,52]. In our study, among patients with profound USNHL, only the two children who had a cochlear implant presented high scores in all audiological evaluations, confirming that cochlear implantation, when not contraindicated, should be the treatment of choice for the pediatric population with SSD [18,43].

Despite the importance of these major findings, this study has several limitations. First, the study is retrospective and, because it is based on a review of medical reports and charts that were not originally intended for research data collection, some information is missing (e.g., there are no data on the mean absolute localization error, use of frequency modulation systems, detailed description of GJB2 pathogenic variants, questionnaires on children's quality of life, children's speech-language skills, and any speech-language therapy sessions after hearing aid fitting). Furthermore, a relatively small number of children with USNHL were evaluated in a single hospital, thus limiting the generalizability of the results. In particular, only two children had a unilateral cochlear implant. Further studies, possibly prospective, conducted across multiple centers and involving a large cohort of pediatric patients with USNHL, are necessary in order to substantiate our findings. Potential future expansions of this research might include the following: (1) an evaluation of long-term audiological and vestibular outcomes in children with USNHL; (2) an assessment of sound localization, speech perception in both quiet and noisy environments, and speech-language skills before and several months after hearing device fitting; and (3) translation and adaptation into the Italian language of some existing English questionnaires aimed

at estimating children's listening abilities in different everyday situations, such as the Children's Home Inventory for Listening Difficulties (C.H.I.L.D.) [53].

5. Conclusions

The present study showed that a high percentage of patients with USNHL were diagnosed after the age of 2 years, confirming the importance of performing regular surveillance of developmental milestones and auditory skills in all children, regardless of newborn hearing screening outcome. Although all children underwent extensive diagnostic evaluations, in many cases, the etiology of USNHL was idiopathic. The management of children with USNHL should include the accurate and timely diagnosis of HL, appropriate selection of amplification devices, verification of the fitted devices, and long-term audiological follow-up. Cochlear implantation should be considered a viable treatment option for children with SSD, especially those at risk of hearing deterioration in the contralateral ear. More efforts should be made to raise awareness among professionals and parents about the negative consequences of uncorrected USNHL. Rigorous and updated national and international guidelines for the management of pediatric UHL are a priority.

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Institutional Review Board Statement: This retrospective study was conducted in accordance with the World Medical Association's Declaration of Helsinki and received approval from the ethics committee (Area 2 Milano) of the Fondazione IRCCS Ca' Granda Ospedale Maggiore Policlinico (approval code: 468; approval date: 2 January 2019).

Informed Consent Statement: Informed consent was obtained by the children's parents.

Data Availability Statement: The data presented in this study are available upon request from the corresponding author.

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Article

Audiophonologopedic Telerehabilitation: Advantages and Disadvantages from User Perspectives

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Abstract: Introduction: Technological advancements and the COVID-19 pandemic have highlighted the importance of digital tools for patient care and rehabilitation. This study explores user perspectives on telerehabilitation, comparing it to traditional methods and identifying criteria for determining its suitability for different patients and clinical conditions. Methods: This study was carried out during the period of May–September 2021. Questionnaires were administered to 48 users in rehabilitation for audiophonologopedic and neurodevelopmental disorders in three rehabilitation centres in central Italy. Results: The user responses predominantly emphasize the benefits of time saving (68.75%) and cost-efficiency (37.5%), specifically regarding time saving due to travel and expenses incurred to go to where the therapy is carried out. The disadvantages include increased distraction (60.42%) in following the instructions remotely and logistic problems (39.58%). Patients with hearing loss were subjected to a larger number of telerehabilitation sessions, positively rating this alternative method. Patients with speech and language delay and autism spectrum disorder (ASD) prefer traditional treatment. Discussion: This study reveals a favourable perception of telerehabilitation as a therapy approach to be regarded as a supplement or temporary option to the irreplaceable face-to-face one. More research, as well as a larger sample sizes, will be useful to increase the significance of the correlations reported in this study.

Keywords: e-health; telemedicine; telerehabilitation; COVID-19; neurodevelopmental disorders; hearing loss; user perspective

1. Introduction

Telemedicine, now defined as telehealth or e-health [1], is becoming a popular alternative method to provide healthcare services supported by Information and Communication Technologies (ICTs). It is especially useful when a direct patient–healthcare provider relationship is not possible [2,3], with the aim of ensuring hospital–territory continuity, care accessibility, and cost savings, while maintaining service quality and universality [4]. The large number of papers and research works on telemedicine highlight an increasing interest in this field [5], especially after its wide use during the COVID-19 pandemic, when services were limited by lockdown measures imposed by Governments to contain the spread of SARS-CoV-2 infection. Due to the lockdown, patients experienced delays, discontinuances, and cancellations to their treatments, weakening their effectiveness [6]. As a result, the pandemic created the need for new health services by enhancing remote performances and

developing national guidelines for telemedicine and telerehabilitation [7,8]. Rehabilitation services found new opportunities through digitisation, which led to the telerehabilitation, a specialized branch of telemedicine for rehabilitation, consultation, and monitoring activities. Telerehabilitation has different applications, including motor and cognitive rehabilitation, as well as occupational, communication, deglutition, behavioural, cardiological, and respiratory disorders. Furthermore, telerehabilitation can be used for the prescription and evaluation of aids, orthoses, and prostheses. Telerehabilitation required technological supports (tablet, smartphone, computer), as well as a stable and high-quality Internet connection and a therapy-dedicated station. The sudden interruption of traditional rehabilitation approaches during the pandemic particularly hit those patients with chronic diseases, rare diseases, neurodevelopmental and psychiatric disorders, psychophysical disabilities, and patients who need medium- and long-term continuity of care [8]. The guidelines provided by the Italian Ministry of Health [8] were characterized by measures implemented towards mental health protection and vulnerable subjects, particularly pediatric ones [9–11]. The purpose of this medical and political act goes far beyond the reduction in the consequences of a specific disorder, but aims to develop the adaptive function of the subject, to realize self-sufficiency, and to improve the quality of life, sociality, and work contribution [12]. Therefore, any problem in accessing a rehabilitation treatment means irreversibly jeopardizing psychophysical development, defining an irreversible disability and the failure to adapt in society with all the related consequences [13]. Because of its multidisciplinary characteristic, telerehabilitation is particularly well-suited for the delivery of the audiophonologopedic therapy [14–25]. The chance of a “tailored” therapy based on the patient’s needs and preferences, combined with the flexibility of a remote therapeutic approach, contributes to the effectiveness of the rehabilitation process [26,27]. Guidelines recommend the evaluation of the available resources related to the territory, family, and social environment, as well as the potential obstacles to the treatment, in order to adjust the therapeutic plans to empower the appropriateness of the intervention with the goal to achieve positive outcomes. The evaluation of functional and care outcomes is carried out to monitor the rehabilitation progress together with the user and/or caregiver’s satisfaction and treatment perception (e.g., Patient Reported Outcome Measure) [8]. Few data are currently available on the satisfaction perceived by telerehabilitation actors (parents, users, clinicians, and therapists). Scientific studies are available in the literature that highlight the inconsistent outcomes of telerehabilitation, generating scepticism also among parents regarding the use of a remote assistance support [28,29]. The aim of this study on audiophonologopedic telerehabilitation is to collect scientific data regarding the experience reported by the users of this tool and which benefits and drawbacks were emphasized [30]. Based on the results of this study, together with those provided by further research, we can better outline specific fields of intervention with telerehabilitation: when it is recommended, when it is not recommended, and even when it can give rise to negative outcomes, always considering the clinical condition and the age of the patient, as well as the available resources. So far, the therapeutic options developed with the telerehabilitation have proven to be feasible both from an organizational and economic point of view, but only a prolonged experience over time supported by scientific evidence will confirm the full clinical validity of telerehabilitation and its actual incorporation into clinical practise as a supplement to the traditional method.

2. Materials and Methods

The reference population of this study, conducted from May to September 2021, includes patients treated in three rehabilitation centres in central Italy. These are the patients whose parents have accepted to participate in this study and to fill out the scheduled questionnaire. The heterogeneity of the population included in the study is due to the inclusion criteria: no limits for age and type of pathology were set with the result of eight different pathological frameworks. We did not use strict exclusion criteria because we collected preliminary data to evaluate as many patients as possible, as there are not

many similar studies already performed using user perspectives. All the patients carried out speech therapy aimed at the re-education or education of expressive, receptive, and communication language. The subjective questionnaire was provided to the users who received remote speech rehabilitation during the COVID-19 pandemic. The users were required to respond anonymously, and, in the case of minors, young children, or patients with a clinical condition that prevented a complete comprehension, the questionnaire was completed in the presence of a parent or caregiver, who eventually reported their answers. Participation was on a voluntary basis, and every user signed the informed consent form to the processing of anonymous data for the research project, approved by the Internal Review Board (Prot. N. 41818, 5 April 2022, University of L'Aquila). A questionnaire of 17 questions was developed, which provides general user information (age, origin, and pathology), investigates rehabilitation delivery methods (synchronous or asynchronous, direct, or indirect) and the type of platforms used, highlights the user preference (face-to-face or remote therapy), and assesses the perception of improvements achieved after telerehabilitation (Supplementary Material). After a brief explanation of the study and research objectives, the questionnaire and informed consent were supplied in paper form; users completed both and placed the two papers into separate boxes. Some participants electronically signed the consents and completed the survey providing responses through email or WhatsApp/Telegram. Others answered the structured interview-style questionnaire in the presence of the speech therapist and parent/caregiver. After data collection, Stata 15 software(version:15.1) was used (Statistics/Data Analysis TA USA, Stata Corp LP, College Station, TX, USA). Stratification by pathology, treatment method, improvements, distractions, and time saving was carried out to evaluate the distribution of frequency. A χ^2 test was used to determine whether or not there was a statistically significant difference between the groups. A p value ≤ 0.05 was considered significant.

3. Results

The reference sample for the analysis consists of 48 users (42 children and 6 adults; mean of users age 11.8 years old; SD 10.7). The pathologies of the participants are reported in Table 1.

Table 1. Pathologies reported by the participants. ASD: autism spectrum disorder; ADHD: attention deficit hyperactivity disorder.

Pathologies	%
hearing loss	29.17%
ASD	18.75%
speech and language delay	16.67%
specific learning disorder	14.8%
intellectual disability	14.58%
ADHD	4.17%
cleft palate	2.08%

It is important to note that all users began rehabilitation treatment prior to the COVID-19 emergency, and none claimed that they would utilize this alternative approach until 2020.

When the reference centre's multidisciplinary team proposed conducting remote sessions, 76% of users felt reassured about the idea of not interrupting treatment; instead, 19.57% were sceptical about this method.

Telerehabilitation requires the possession of Internet connection and digital devices, such as computers, tablets, and mobile phones: only a small group of patients (10.42%) did not have the essential tools to access to remote rehabilitation. We did not evidence a significant difference in the results based on the type of device used for telerehabilitation because all the users utilized computers. The platforms, dedicated web sites, used and the messaging and video calling apps used are reported in Table 2.

Table 2. Platforms, dedicated web sites, and messaging and video calling apps used by the participants.

	%
trainingcognitivo.it (accessed on 24 August 2024).	8.70
Both Skype (iOS 8.123.0.203) and Zoom (6.1.11.45504)	6.52
Only Skype	6.52
Both trainingcognitivo.it and wordwall.net (accessed on 24 August 2024)	4.35
Only wordwall.net (accessed on 24 August 2024).	2.17
WhatsApp (Version 22.13.74)	2.17
Not specified	19.57

Another fundamental element to consider for the success of telerehabilitation is the autonomy of the user during the session. Few patients did not need to seek assistance (12.50%) during the session. The majority of patients needed help from a parent/caregiver in every session (37.50%) or only the first time (39.58%). In other cases, a speech therapist guided the user in case of need (10.42%), asking for the help of the parents by phone or during the session of telerehabilitation.

Speech therapy involved individual sessions (72.34%) or mixed ones, also including co-presence sessions (27.66%). A co-presence session was composed by more users together in a session, where everyone had their own speech therapist.

Regarding the number of weekly telerehabilitation sessions, the users mainly participated once a week (34.04%) and twice a week (55.20%).

The importance of therapy continuity is particularly underscored in replies relating to session frequency: more than 67% of patients maintained the same involvement, while 20% reported increased attendance with telerehabilitation.

Although telerehabilitation has been expressly requested by patients in the later phases of the COVID emergency, it is essential to point out that the majority of users believe that face-to-face treatment is more helpful for the management of various pathologies [Table 3].

Table 3. Users' response percentages related to the pathology and treatment methods. ASD: autism spectrum disorder; ADHD: attention deficit hyperactivity disorder.

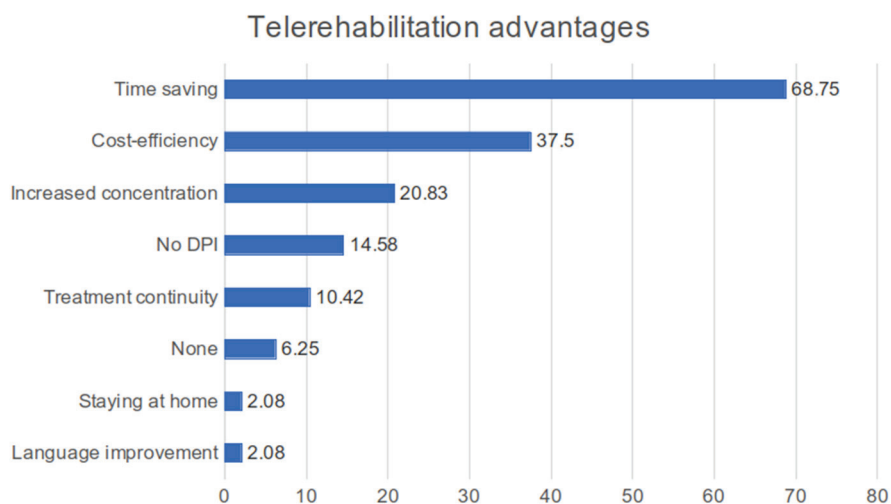
KERRYPNX			Treatment Method Which is Perceived as the Best						
■ Pathology			Telerehabilitation		In Person		Both		
■ n	Total		n	(%)	n	(%)	n	(%)	<i>p</i> Value
■ Speech and language delay	8		0	(0.00)	8	(100.00)	0	(0.00)	0.492
■ Specific learning disorder	7		1	(14.29)	6	(85.71)	0	(0.00)	
■ Intellectual disability	7		1	(14.29)	5	(71.43)	1	(14.29)	
■ ASD	9		0	(0.00)	8	(88.89)	1	(11.11)	
■ Hearing loss	14		3	(21.43)	7	(50.00)	4	(28.57)	
■ ADHD	2		0	(0.00)	2	(100.00)	0	(0.00)	
■ Cleft palate	1		0	(0.00)	1	(100.00)	0	(0.00)	

Subjects with an intellectual disability or ASD did not perceive improvements via telerehabilitation, which was seen as more of a maintenance of the obtained results. Patients with hearing loss, instead, have described perceived improvements, and some have even reported better results with remote treatment [Table 4].

Table 4. Users' response percentages related to the pathology and noted improvements. ASD: autism spectrum disorder; ADHD: attention deficit hyperactivity disorder.

Have You Perceived Any Improvements?							
	No, I Have Not. It Was Useful as Maintenance		Yes, I Have Perceived the Same Grade of Improvements		Yes, I Have Perceived Better Improvements with Telerehabilitation		
■ Pathology	n	(%)	n	(%)	n	(%)	<i>p</i> Value
■ Speech and language delay	2	(25.00)	5	(62.50)	1	(12.50)	0.246
■ Specific learning disorder	4	(57.14)	3	(42.86)	0	(0.00)	
■ Intellectual disability	5	(71.43)	2	(28.57)	0	(0.00)	
■ ASD	5	(62.50)	2	(25.00)	1	(12.50)	
■ Hearing loss	1	(7.14)	11	(78.57)	2	(14.29)	
■ ADHD	1	(50.00)	1	(50.00)	0	(0.00)	
■ Cleft palate	0	(0.00)	1	(100.00)	0	(0.00)	

Those who filled out the questionnaire were asked to express the advantages they perceived with telerehabilitation. The main advantages are time saving (68.75%), cost-efficiency (37.5%), and increased concentration (20.83%) [Figure 1].

**Figure 1.** Histogram of answer percentages about telerehabilitation advantages according to users' points of view. DPI: individual protection devices.

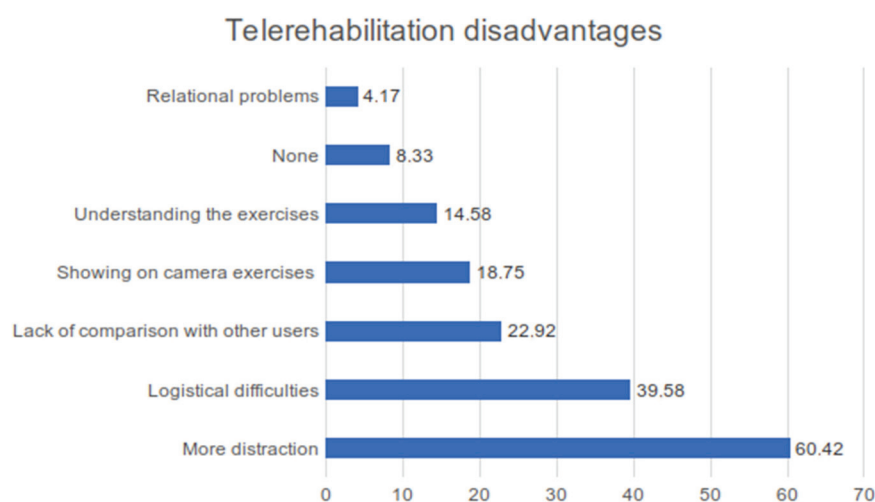
The first two are related to a more organizational–management aspect of rehabilitation, which reflects the caregiver's point of view: remote treatment allows you to avoid long journeys to reach the rehabilitation centre, a non-secondary factor in cities like Rome, and allows you to save on the cost of fuel, tolls, and transportation tickets.

Statistically, significant associations have been identified to confirm this: almost all patients with hearing loss (92.86%; p -value = 0.020), those who prefer telerehabilitation (100%; p -value = 0.041), and also users who rather use the traditional method (59.46%, p -value = 0.041) consider time saving an advantage. In general, all the patients interviewed recognize that time saving in remote therapy is essential, although this is not considered by all to be the best way of providing treatment for their pathology (p = 0.039). Furthermore, 72.97% of patients claim to be distracted during face-to-face therapy and 80.00% of them claim to maintain concentration with remote therapy (p = 0.005) (Table 5)

Table 5. Percentages of time saving and distraction reported by patients with telerehabilitation, in person and both.

Treatment Perceived as the Best	Time Saving		No Time Saving		<i>p</i> Value	Distraction		No Distraction		<i>p</i> Value
	n	%	n	%		n	%	n	%	
Telerehabilitation	5	100.00	0	0.00	0.039	1	20.00	4	80.00	0.005
In person	22	59.46	15	40.54		27	72.97	10	27.03	
Both	6	100.00	0	0.00		1	16.67	5	83.33	

Logistical difficulties (39.58%) and more distraction (60.42%) are, according to the users, the main telerehabilitation disadvantages [Figure 2].

**Figure 2.** Histogram of answer percentages about telerehabilitation disadvantages according to users' points of view.

More distraction has a statistically significant association with ASD (88.89%; p -value = 0.05) and intellectual disability (100%; p -value = 0.020). So, based on these statistical data, telerehabilitation is not particularly appropriate for these disorders. In total, 64.29% of patients with hearing loss report an absence of distractions.

There is a statistically significant association ($p = 0.033$) between distraction and pathologies. However, this association is not statistically significant ($p = 0.112$) between time saving and pathologies (Table 6).

Table 6. Percentages of time saving and distraction reported by patients in telerehabilitation.

Pathology	Distraction		No Distraction		<i>p</i> Value	Time Saving		No Time Saving		<i>p</i> Value
	n	(%)	n	(%)		n	(%)	n	(%)	
Speech and language delay	5	62.50	3	37.50	0.033	3	37.50	5	62.50	0.112
Specific learning disorder	3	42.86	4	57.14		4	57.14	3	42.86	
Intellectual disability	7	100.00	0	0.00		1	14.29	6	85.71	
ASD	8	88.89	1	11.11		4	44.44	5	55.56	
Hearing loss	5	35.71	9	64.29		13	92.86	1	7.14	
ADHD	1	50	1	50		1	50.00	1	50.00	
Cleft palate	0	0.0	1	100.00		1	100.00	0	0.00	

The analysis shows a significant association between the distraction variable and three individual pathologies: intellectual disability ($p = 0.033$), ASD ($p = 0.05$), and hearing loss ($p = 0.025$) (Table 7).

Table 7. Significant association between the distraction variable and intellectual disability, ASD, and hearing loss.

Pathology	Distraction		No Distraction		<i>p</i> Value
	n	(%)	n	(%)	
Speech and language delay	5	62.50	3	37.50	0.895
Specific learning disorder	3	42.86	4	57.14	0.304
Intellectual disability	7	100.00	0	0.00	0.033
ASD	8	88.89	1	11.11	0.050
Hearing loss	5	35.71	9	64.29	0.025
ADHD	1	50	1	50	-
Cleft palate	0	0.0	1	100.00	-

4. Discussion

The technological progress, together with the large number of patients increasingly accountable for their own health, with an active part in the decision-making process for the assessment of the therapeutic programme, allowed for a quick upward evolution in the use of digital technology for rehabilitation. Furthermore, the need to guarantee an adequate level of assistance during the pandemic forced the use of telerehabilitation in clinical practise, both in Italy and abroad. This surge was also aided by the progressive interest in the scientific literature on this topic and the effectiveness of telemedicine therapies and telerehabilitation [31,32]. However, these studies all have the limitation of research being conducted with a specific focus on the qualitative values and not on the quantitative ones, resulting in a loss of statistical strength. This study aims to collect and examine the advantages and disadvantages of telerehabilitation from the user's point of view, a topic that has received little attention in the scientific literature.

The same Italian guidelines on remote rehabilitation consider the patient's opinion to be critical to better evaluate the indications for an effective therapy conducted in a remote mode with improved multidisciplinary management.

The sample consists of patients from rehabilitation centres in central Italy: 48 users, or their parents/caregivers, who completed the questionnaire. The patients were treated for hearing loss, ASD, intellectual disability, speech and language delay, specific learning disorders, ADHD, and cleft palate.

All participants started with face-to-face treatment before the pandemic, and they had never used telerehabilitation before, despite the fact that it was already available. Since the implementation of the lockdown measures, the use of a remote mode therapy has been recommended to prevent interruption to or discontinuity in rehabilitation.

Telerehabilitation was supported by nearly three-quarters of the patients who were concerned about the reduced frequency or interruption of the therapy. However, in a small percentage of cases, doubts and worries were expressed about this new mode of intervention. Fortunately, almost every family already had the technological support (tablet, smartphone, computer) for telerehabilitation, as well as a stable and high-quality Internet connection and a therapy-dedicated station. Despite the rapid digitalisation of society, some interviewees said that they did not have the appropriate devices at first and thus had to obtain them quickly. This highlights the presence of population groups that do not have easy access to technological progress due to economic constraints.

After the first emergency phase (May 2020), not all the patients returned to their usual home, and some of them wanted to continue the telerehabilitation for their own needs or, always in agreement with the therapist, opted for a combination of the two modalities from remote and face-to-face treatment. The adhesion to the therapy sessions was stable for many users, decreased only for a small number of patients, and increased for nearly a fifth of the interviewees. This can be explained by the better organizational ease with which the remote modality was used, as it was also used for work and educational purposes during the pandemic.

We attempted to establish the relationship between the modality of the therapeutic sessions and the disease, but the current amount of data does not allow us to consider them as significant (p -value > 0.05). However, we can find some correlations showing that individual sessions are carried out mostly by patients with speech and language delay and hearing loss. Patients with hypoacusis have performed more sessions of both types, individual and co-presence.

The collaboration between the professionals and the families involved in the treatment was essential, especially during the early phases of telerehabilitation requiring some training and adaptation. Many users stated that they needed assistance and were initially guided by a speech therapist.

Another key component to examine to evaluate telerehabilitation is the perception of specific improvements. More than half of the interviewees believe that the progress achieved was similar in both modalities, and many of them think that remote rehabilitation can be as useful as valuable for maintenance and to consolidate the results obtained with the therapy.

With the limitations of the current data, it should be considered that despite the positive qualities noted with telerehabilitation, face-to-face rehabilitation is seen as the most effective method, satisfying the preferences of patients with ASD and speech and language delay. For the patients with hearing impairment, remote rehabilitation can be an instrument that allows them to achieve similar results and sometimes results superior to those obtained in person. Although statistical significance for some correlations should be studied with a larger sample size, we can still consider positively the results obtained during telerehabilitation, as well as the fact that was not to interrupt a non-delayable treatment. The most obvious benefits are time saving and economic convenience, related to the organizational aspect of rehabilitation. The understanding of these advantages must be linked to the fact that the majority of the interviewed users were from Rome and its suburbs, and with telerehabilitation they would save time otherwise wasted in traffic and travelling large distances. It is also important to consider the economic benefit for a reduced use of cars, as well as the use of public transportation and the cost of the ticket. There is a statistically significant correlation between time saving and hearing loss pathology. Furthermore, this advantage is recognized by both individuals who believe that telerehabilitation is the most effective treatment for their condition and those who prefer traditional treatment. A minority of users and caregivers, observing patients' behaviour, noticed an increased concentration with the remote mode. There is a significant correlation for this advantage between those who did not see the increase in concentration and the users who saw telerehabilitation as a maintenance treatment only.

This allows us to speculate about reported improvements and a lack of attention, which is supported by another statistically significant correlation between observed improvements and one of the perceived disadvantages, more distraction during telerehabilitation sessions. As a result, not seeing improvements can be attributed to the distraction caused by distance and the digital barriers. This disadvantage is also associated with pathologies such as intellectual disability and ASD, both of which are difficult to handle by the therapist remotely. Other drawbacks identified by users include logistical issues related to the stability of Internet connection and the availability of a station dedicated to therapy, difficulty in understanding the exercises to be performed, and a lack of the social aspect that is normally guaranteed by attendance at centres. The emotional side was partially compensated by the ability to establish eye contact via digital means. Less easy, however, was the total home management of patients with complex clinical situations, in which the caregiver often received little assistance [33,34]. Verma et al. evaluated 27 unilateral pediatric cochlear implantees in the age range of 2–11 years, divided into two groups based on the therapy modality, telerehabilitation and face-to-face. The study indicated that the conventional method of the speech–language and auditory rehabilitation was far better compared to the telerehabilitation services with the WhatsApp video calling platform, as majority of the population were from lower socioeconomic backgrounds with

poor educational backgrounds. The limitations reported with the teletherapy mode were connectivity issues, poor sound quality, and poor visibility [35].

4.1. Limitations

The small sample size of this study may affect the representativeness and statistical significance of the results. In future research, we will increase the sample size to improve the reliability of the results. The current research mainly focuses on patients with hearing loss, language delay, and autism spectrum disorder. We will include more types of rehabilitation patients in future research to obtain more comprehensive conclusions. We will also introduce future studies in which different age groups are separated, considering the subjectivity of the proposed questionnaires. It would also be very interesting to compare the results obtained by groups of patients of the same age and with the same pathology, who performed the rehabilitation course in person versus those who performed part of it in telerehabilitation.

4.2. Follow Up

The use of telerehabilitation did not stop in the Audiophonologopedic Center of Rome with the end of the pandemic emergency, but it continued. Currently, three users are being followed with exclusive telerehabilitation, including two girls (15.8 and 14.1 years old) with sensorineural hearing loss, wearing cochlear implants, and a child (7.11 years old) suffering from a neoplastic disease being treated at a hospital and, therefore, in a frail condition not compatible with in-person therapy. The reasons, in the case of the two girls, are to be ascribed to the distance of their homes from the centre's headquarters and the economic difficulties of both families that mean it would not be possible to accompany them, losing hours of work and forcing them to spend a greater amount of energy taken away from studying and socialization with peers. For the remaining 193 users affected by the types of pathologies illustrated in the materials and methods, telerehabilitation is used in a systematic and integrated way in the service delivery processes, with the exception of children with autism spectrum disorder and/or a chronological age of under 6 years, as in the following cases:

- To encourage the recovery of therapies not provided, even at times not foreseen by the rehabilitation project;
- To allow them to benefit from the treatment at the scheduled time in the event of an obstacle to attendance at the centre due to unexpected family reasons;
- To guarantee the treatment in case of mild ailments/convalescence phases, which do not allow attendance in person, but are compatible with rehabilitation work.

In all cases, the telerehabilitation option provided greater flexibility and guaranteed the continuity of work, reducing the number of absences with both organizational advantages for the structure and clinical–rehabilitative ones for the users. A new questionnaire plans to record the advantages and disadvantages of the hybrid use of telerehabilitation.

A study about telerehabilitation in families with children with neurodevelopmental disorders during the lockdown caused by the COVID-19 showed high levels of participation and satisfaction. The authors suggested sharing the resources for their applicability in other countries where families have similar needs conditioned by COVID-19 [36].

5. Conclusions

This study is hampered in part by the small number of participants, yet this does not prevent us from finding interesting observations.

Remote rehabilitation has undoubtedly played a functional role in the evaluated experience, ensuring treatment continuity and avoiding the unsuitable psychophysical development of the children. Further studies may assess its actual future relevance as an integration method in the rehabilitation path or as an alternative in case of need, especially as this field of study represents a research stimulus for the clinical, rehabilitation, and technological sectors.

Supplementary Materials: The following supporting information can be downloaded at: <https://www.mdpi.com/article/10.3390/children11091073/s1>, Supplementary File S1: USER QUESTIONNAIRE.

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Review

Music-Based Interventions in Childhood Hearing Loss: A Comprehensive Narrative Review

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Highlights

What are the main findings?

- Children with hearing loss show reduced pitch and timbre perception.
- Musical training can improve auditory, linguistic, and cognitive skills.
- Music therapy studies are few, but available evidence suggests benefits that extend beyond auditory perception to emotional, social, and behavioral domains

What are the implications of the main findings?

- Music-based interventions may support broader communicative development by leveraging rhythmic strengths and enhancing auditory processing.
- Expanding and standardizing music therapy approaches could improve holistic care for children using cochlear implants or hearing aids.

Abstract

Background/Objectives: Childhood hearing loss (HL) affects auditory, linguistic, and social development. Alongside conventional rehabilitation, music-based interventions have gained increasing attention for their potential to support both auditory and non-auditory domains. This narrative review aims to summarize current evidence on the use of music-based interventions in children with HL. **Methods:** A narrative review of the literature was conducted, examining studies involving pediatric cochlear implant or hearing aid users. Publications were categorized into three main areas: musical auditory perception, musical training, and music therapy. **Results:** Studies on musical auditory perception demonstrate persistent limitations in pitch and timbre perception in children with HL, while rhythmic abilities appear relatively preserved. Musical training interventions, particularly those targeting rhythm, have been associated with improvements in auditory perception, linguistic processing, and selected cognitive skills, although parental involvement and long-term designs remain limited. Existing literature on music therapy is scarce but suggests potential benefits extending beyond auditory skills to emotional regulation, social interaction, and quality of life. **Conclusions:** Music-based interventions represent a promising complementary approach in pediatric hearing rehabilitation. While musical training is more widely studied, music therapy is still underrepresented despite its holistic focus. Further structured studies are needed to define standardized protocols and outcome measures for music therapy in children with HL.

Keywords: musical auditory perception; musical training; music therapy; cochlear implants; hearing aids; hearing loss

1. Introduction

Childhood hearing loss (HL) can markedly affect a child's communication abilities, emotional growth, and social participation [1]. Limited auditory input often results in delays in speech development, difficulties in language comprehension, and reduced engagement with peers. These challenges may extend beyond communication, influencing academic performance, self-esteem, and overall quality of life [2]. Early, well-structured strategies remain essential to address the developmental vulnerabilities associated with pediatric HL. Music therapy has increasingly been recognized as a supportive intervention that addresses both auditory and non-auditory developmental domains [3]. Through structured musical experiences such as rhythmic activities, movement, singing, and instrumental interaction, music therapy stimulates multiple sensory pathways and fosters meaningful engagement [4]. Even in cases of moderate-to-profound HL, music can be perceived through vibrations, visual information, and tactile cues, allowing children to actively participate and express themselves creatively [5]. In addition to enhancing auditory awareness, music therapy supports emotional regulation, social interaction, and cognitive development. Musical activities promote attention, imitation, and turn-taking, which are skills closely associated with language acquisition [6]. Moreover, the enjoyable and motivating qualities of music may reduce anxiety and strengthen therapeutic relationships [7].

The present work provides a narrative review of the literature on the use of music-based interventions in pediatric patients with HL, including both cochlear implant (CI) and hearing aid (HA) users. The aim of this review is to offer an overview of the clinical interventions and practical experiences described to date on this topic. A comprehensive search was performed in the electronic databases PubMed/MEDLINE, Scopus, and Web of Science, including all publications available up to February 2026. The search strategy combined Medical Subject Headings (MeSH) and free-text terms related to HL and music-based interventions, using combinations of the following keywords: "hearing loss", "cochlear implant", "hearing aid", "children", "pediatric", "auditory perception", "music", "musical training", and "music therapy". Boolean operators (AND/OR) were applied to refine the search and ensure broad coverage of relevant studies. Studies were considered eligible if they involved pediatric populations younger than 14 years with HL using CI and/or HA and investigated music-based interventions—musical auditory perception, musical training, or music therapy—provided that they reported original data. Research conducted exclusively in adult populations, articles not focused on music-related interventions, and non-original contributions such as editorials, commentaries, or conference abstracts lacking sufficient methodological detail were excluded. Specifically, the selection process was conducted in three stages. First, titles and abstracts identified through the database search were screened to exclude clearly irrelevant articles. Subsequently, the full texts of potentially eligible studies were retrieved and assessed for inclusion based on the predefined criteria. Finally, the reference lists of the included articles were manually screened to identify additional relevant studies. Given the narrative nature of this review and the heterogeneity of the included studies, no formal quality assessment or risk-of-bias tool was systematically applied; however, key methodological aspects, such as sample size, study design, and the presence of control groups, were taken into account during data interpretation. Overall, this work provides a narrative overview of the available literature

rather than a formal systematic review, as the currently published studies are relatively few and differ substantially in terms of study design, interventions, and outcome measures. To facilitate a coherent structure, the analysis is organized into three main sections: musical auditory perception, musical training, and music therapy.

It is important to clarify the distinction between musical training and music therapy, as these terms are often used interchangeably despite referring to conceptually different approaches. Although they differ in several key aspects, the two modalities also share certain objectives and methodological elements. According to the World Federation of Music Therapy, music therapy is defined as “*the professional use of music and its elements as an intervention in medical, educational and everyday environments with individuals, groups, families or communities who seek to optimize their quality of life and improve their physical, social, communicative, emotional, intellectual and spiritual health and wellbeing. Research, practice, education and clinical training in music therapy are based on professional standards according to cultural, social, and political contexts*” [8]. From this definition, two key elements emerge that distinguish music therapy from other interventions involving sound and music: the presence of a specifically trained professional and the broad range of objectives related to the individual’s overall wellbeing, encompassing cognitive, emotional, behavioral, communicative, and interpersonal domains [9]. Interventions defined as musical training, by contrast, may be delivered by personnel without specific therapeutic training, sometimes by family members themselves or by teachers, and pursue objectives more narrowly related to the auditory domain, such as improvements in sound perception and in the discrimination of sounds and speech [10]. The use of programs or applications offering musical exercises and games falls predominantly within musical training; however, these tools may occasionally also be used in music therapy as a complement to other activities. Conversely, experiences defined as musical training often include playful auditory–musical activities that are also widely used in music therapy, such as singing, music listening, sound production with musical instruments, and exercises related to the perception of musical parameters and musical emotion [11]. As a matter of fact, the boundary between musical training and music therapy remains partially fluid in practice, as both approaches may employ similar techniques and, in some cases, pursue overlapping functional outcomes. This convergence reflects the shared use of music as a multisensory and relational medium, while differing primarily in therapeutic intent and professional framework. Accordingly, in the present review, an operational distinction was adopted based on the leading aim of the intervention and the clinical context in which it was delivered, in order to ensure conceptual clarity while acknowledging the existence of a continuum between these modalities. The key concepts and definitions of the terms used throughout this review are presented in Table 1.

Table 1. Key concepts and definitions related to music and hearing loss.

Term	Definition	Functional Role
Cochlear implant	A surgically implanted device that bypasses damaged structures of the cochlea and delivers patterned electrical stimulation directly to the auditory nerve.	To restore access to sound and enable speech and music perception within the constraints of current technology.
Hearing aid	A wearable device that amplifies and shapes the acoustic signal for individuals with HL.	To enhance audibility, supporting the perception of environmental sounds, speech, and music.

Table 1. Cont.

Term	Definition	Functional Role
Musical auditory perception	The ability to process, analyze, remember, and predict the acoustic patterns conveyed by music, including temporal elements such as rhythm and spectral features such as pitch and timbre.	To support auditory, cognitive, and linguistic development by engaging attention, memory, and sensorimotor integration.
Music-based interventions	Structured use of musical activities within therapeutic, educational, or rehabilitative frameworks.	To modulate auditory processing, linguistic development, cognitive functions, emotional regulation, and social participation.
Music therapy	A clinical discipline in which trained and credentialed music therapists use structured musical experiences.	To achieve individualized developmental, communicative, emotional, behavioral, and psychosocial health outcomes.
Musical training	Systematic, goal-directed instruction in musical activities, typically without a therapeutic mandate and often delivered by non-clinical personnel.	To strengthen musical and auditory competencies, such as auditory discrimination, fine motor skills, and multisensory integration.
Pediatric hearing rehabilitation	A set of coordinated clinical and educational interventions designed for children with HL, involving auditory devices, therapeutic strategies, and communication support approaches.	To improve language, learning, and social participation through optimized auditory access and communication support.

HL = Hearing loss.

2. Musical Auditory Perception in Children with Hearing Loss

In the articles addressing musical auditory perception, the primary focus is the evaluation of how children with HL perceive music and its constituent parameters. In the literature, musical auditory perception is described as *“the individuals’ abilities to process, analyze, remember, and predict acoustic patterns, auditory scenic content, and interval relations of syntax and semantics conveyed by music”* [12]. Music represents a primary perceptual stimulus for the human auditory system and is closely related to the early development of auditory and linguistic functions. The brain areas involved in musical and linguistic perception are anatomically adjacent and partially overlapping, suggesting a functional interaction between the two domains [13]. It has been observed that good musical perception is associated with better cognitive and emotional abilities. For this reason, recent research has aimed to improve the musical experience of CI users, while also promoting social integration and an improved quality of life [14]. However, the literature highlights significant limitations in the perception of musical elements in individuals with HL and in CI users. In particular, the temporal components of the acoustic signal (e.g., rhythm) are better encoded and therefore more easily perceived than the spectral and frequency components (such as timbre and pitch), which require a more accurate representation of the sound signal [13]. This occurs due to the reduced precision of spectral transmission of the acoustic signal, which is intrinsic to the electrical stimulation model of CI [15]. The technological limitations of CI result in significant difficulties in musical perception, mainly due to reduced spectral resolution and a limited dynamic range compared with normal hearing [16]. This limitation of CI often affects language development, as the perception of auditory information relies on variations in pitch, amplitude, and duration of speech [17]. Nevertheless, targeted musical training after cochlear implantation can enhance musical perception [18]. Since musical perception is shaped not only by acoustic and technological

factors but also by emotional responses to musical stimuli [16,18,19], several neurobiological and developmental mechanisms have been proposed to explain why music-based activities may support auditory, linguistic, cognitive, and socio-emotional development in children with HL [20–34]. These mechanisms are summarized in Table 2.

Table 2. Potential neurobiological and developmental benefits of music-based interventions in children with hearing loss.

Domain	Potential Mechanisms	Proposed Benefits
Attention and executive functions	Engagement of fronto-temporal and fronto-striatal networks during musical interaction.	Improved attentional control, working memory, and cognitive flexibility.
Auditory cortical plasticity	Repeated exposure to structured sound patterns enhances neural responsiveness within auditory cortical networks.	Improved sound discrimination, pitch perception, and auditory pattern recognition.
Cross-modal sensory integration	Interaction between auditory, motor, and somatosensory neural systems during musical activities.	Improved multisensory integration and support for speech–motor coordination.
Emotional and social processing	Activation of limbic and reward-related neural systems during musical interaction.	Enhanced emotional expression, social engagement, and parent–child interaction.
Language and phonological processing	Overlap between neural circuits for music and speech processing.	Support for phonological awareness and early language development.
Motivation and engagement in rehabilitation	Music-driven reward and dopaminergic modulation of learning circuits.	Increased participation in and adherence to rehabilitative activities.
Temporal processing and rhythmic entrainment	Synchronization of neural oscillations with rhythmic stimuli.	Enhanced speech perception, prosody recognition, and timing of auditory processing.

2.1. Study Samples

Across most of the reviewed studies, sample sizes typically ranged from 27 to 75 children [13–15,17,19]. Two investigations deviated from this range: one study involved a notably small cohort of only five participants [16], while another included a substantially larger sample of 360 children [18]. Regarding methodological design, most studies included control groups that were exposed to the same experimental activities. These control groups were comparable to the HL groups in terms of both participant number and age range [14,16–18]. Only one study adopted a within-group comparison design, examining differences among children with HL without the inclusion of a separate control group [15]. In this case, participants were stratified according to age (older than 6 years versus 6 years or younger) and length of CI use (more than 18 months versus 18 months or less). In two investigations, an additional comparison group was included alongside the HL and normal-hearing cohorts. One study incorporated a group of children with autism spectrum disorder [19], while the other examined a group of children using bimodal auditory stimulation [13].

2.2. Intervention and Assessment Procedures

Studies examining musical parameters primarily assessed abilities such as singing performance, song and melody recognition, timbre discrimination, and responsiveness to rhythmic patterns [18]. In other investigations, participants were presented with pairs of sequential piano tones and were required to judge the pitch relationship between the notes, identifying whether the second tone was the same, higher, or lower than the first [15]. Sev-

eral studies focused on emotional processing through music. These involved listening tasks using prerecorded violin excerpts [19] or selected musical stimuli specifically designed to evoke emotional responses [13]. Participants were then asked to associate the perceived emotions either with corresponding verbal descriptors [19] or with images depicting facial expressions representing those emotions [13]. In relation to speech perception, one study required children with HL to produce reading samples, which were subsequently compared with those of normal-hearing peers. The recordings were evaluated by trained listeners who were blinded to the participants' hearing status to ensure objective assessment [17]. With regard to quality of life and neurophysiological measures, one investigation explored electroencephalographic (EEG) activity while participants viewed a music-based cartoon, providing insight into neural responses to musical stimuli [16]. Assessment approaches varied considerably across studies and included a wide range of standardized tools and measurement techniques. These comprised the Profiling Elements of Prosody in Speech-Communication (PEPS-C), which evaluates receptive and expressive prosodic skills; Diagnostic Analysis of NonVerbal Accuracy 2 (DANVA-2), a measure of nonverbal emotion recognition; and the Contour and Interval subtests of the Montreal Battery of Evaluation of Amusia (MBEA), which assess melodic contour processing and interval discrimination [17]. Additional tools included the Perception of Emotions and Movement in Music test (PEMM), designed to probe emotional and movement-related responses to musical stimuli; the Peabody Picture Vocabulary Test (PPVT) and Expressive Vocabulary Test (EVT), which measure receptive and expressive vocabulary; the Autism Diagnostic Observation Schedule (ADOS), a standardized assessment of social-communication behaviors; and the Clinical Evaluation of Language Fundamentals (CELF), a comprehensive measure of language abilities [19]. Auditory and music-related outcomes were captured using the Category of Auditory Performance (CAP), a functional hearing scale; the Speech Intelligibility Rating (SIR), which evaluates the clarity of speech production; the Musicality Rating Scale (MRS), a clinician-reported measure of musical behaviors; and the Subjective Assessment of Music Enjoyment (SAME), which captures listener-reported enjoyment and engagement with music [14]. Additional methodologies included EEG rhythm monitoring [16] and the "Musical Ears Evaluation Form for Professionals" [18], a structured tool for documenting musical perceptual abilities in clinical or educational settings.

2.3. Summary of Evidence

The evidence emerging from these studies consistently supports the effectiveness of music in therapeutic approaches for children with CI, as greater exposure to music and longer duration of CI use are associated with better musical auditory perception [14,15]. Furthermore, improvements in musical perception appear to play a meaningful role in alleviating language-related difficulties in children with HL, reflecting underlying neuroplastic adaptations associated with CI use [12]. Such changes highlight the potential of music-based approaches to support broader communicative development. Despite these positive effects, children with CI generally demonstrate lower performance in the perception of pitch, timbre, and melody, as well as in the recognition of musical emotions, particularly those relying on spectral sound information [14,19]. In addition, evidence suggests that music may be perceived as less enjoyable by children with CI compared to their normal-hearing peers [16], and that these children may experience greater difficulties with prosodic aspects of speech [17]. Importantly, enhancing musical abilities in children with HL has been linked to improvements in overall quality of life. Gains in musical skills may be accompanied by advances in auditory-verbal abilities and pitch discrimination, consistent with principles of neural plasticity. These findings underscore the importance of promoting

musical engagement in children with CI through the identification and implementation of effective musical interventions and targeted auditory perception training [18].

3. Musical Training in Children with Hearing Loss

Among the different intervention approaches designed to enhance auditory perception or to compensate for hearing-related limitations, a considerable body of research highlights the use of musical training as an effective means of supporting perceptual, linguistic, and cognitive development in children using CI or HA [15,35–47]. Musical training refers to guided engagement in musical activities, typically delivered in educational or recreational contexts rather than as formal therapy. It utilizes auditory and musical stimuli with the aim of strengthening auditory perception, increasing familiarity with hearing devices, and supporting the use of residual hearing capacities [10]. Across the selected studies, considerable variability was observed in participant age ranges, group structures, levels of parental involvement, and the settings in which training was delivered. These variations largely stem from methodological heterogeneity and differing program objectives.

3.1. Study Samples

The studies on pediatric musical training generally involved relatively small samples, most often ranging between 10 and 30 participants. The majority of participants were of school age, although the age distribution varied considerably across studies and was not always clearly reported. In several investigations, samples included approximately 23–29 participants [15,35–38], with children typically aged between 5 and 11 years; only a few studies also included younger participants aged 1–4 years [35,39]. Other studies enrolled smaller cohorts of 7–18 participants [40–47], and some incorporated control groups to allow comparison with typically hearing (TH) peers or alternative intervention conditions [41,44,46,47]. Overall, the available evidence primarily concerns school-aged children, while research involving younger participants remains limited [40].

In a small number of studies, family members were actively involved in administering exercises at home, highlighting the potential role of the family environment in supporting musical training activities [41,43]. With regard to study design, two main types of control were identified. The first is “between-group” control, used in two studies, in which participants on a waiting list received no intervention during the initial experimental phase. This approach allows for a longitudinal comparison of training effects and, in some cases, also serves as a normative reference [35,41]. The second type is “within-group” control, in which the same participants are exposed over time to different interventions, such as an artistic activity or various musical training tasks, enabling internal comparisons within the group [37,38,44,47].

3.2. Intervention and Assessment Procedures

The intervention modalities were varied, but certain groupings can be identified according to specific criteria. Several studies focused on selected parameters of musical perception, aiming to enhance auditory–musical competencies [15,40,41,43,44]. Specifically, these interventions addressed the recognition, discrimination, and reproduction of rhythms and pitches. Within this category, one study investigated how previous musical education could positively influence the recovery of these abilities [15]. In another study, the intervention was based on musical production through singing, measuring parameters such as pitch and intonation [40]. Other studies focused on linguistic and phonological support through musical training [39,42,45]. In these cases, music was primarily used as a supportive tool for linguistic exercises rather than as an explicitly rehabilitative intervention. Accordingly, the outcomes reported across studies reflect their primary objectives and may not capture the

full range of potential effects. In one project, music therapy combined with musical training primarily aimed at improving the child's psychosocial and relational development [47]. Finally, one study used musical elements as a stimulus to enhance executive functions [37].

3.3. Summary of Evidence

Several studies investigated the effects of musical training targeting auditory perception [15,35,36,38,39,41,46]. Results indicate that training is associated with observable improvements in timbre and pitch perception, and that the duration of musical education is positively correlated with pitch discrimination performance. Effects on speech abilities were not consistently observed, while in home-based programs administered by family members, participants showed improvements in note discrimination, with more pronounced gains in musical skills than in linguistic abilities [41,43]. Studies focusing on rhythm demonstrated positive effects on linguistic abilities, such as improved adaptation and responsiveness in verbal interaction, greater phonological accuracy, and increased sensitivity to temporal regularity. In particular, when phonological production was accompanied by regular rhythmic training, improvements in syntactic processing were observed [42,44,45]. Subjective reports suggested that, beyond gains in language and perceptual abilities, participants also experienced psychosocial benefits, particularly in emotional regulation, interpersonal relationships, and broader aspects of quality of life [46,47]. Studies analyzing musical abilities in the absence of a specific intervention revealed that children with CI exhibit significant difficulties in vocal production with accurate intonation, especially in pitch variation, while rhythmic skills remain relatively preserved [40].

4. Music Therapy in Children with Hearing Loss: Approaches and Evidence

A total of five publications describing music therapy approaches for pediatric CI or HA users were identified [46,48–51]. Three studies described music therapy techniques applicable to this population [48–50]. One study reported a two-part survey assessing healthcare professionals' knowledge of music therapy for children with HL and parents' perceptions of its effectiveness following participation in a music therapy program [51]. The remaining study presented a clinical investigation in which music therapy was delivered as part of multimodal measures [46]. Although the intervention also included structured musical training and app-based home exercises, it is discussed within the music therapy domain due to the central role of a certified music therapist, the group-based interactive format, and the primary focus on psychosocial and quality-of-life outcomes. A summary of the included studies on musical training and music therapy is provided in Table 3.

Table 3. Characteristics and key findings of studies on musical training and music therapy in children with hearing loss.

Study (Author, Year)	Population	Intervention Type	Study Design	Key Outcomes
Chen et al., 2010 [15]	Prelingually deaf children with CI	Musical training	Controlled experimental study (between-group)	Improved pitch perception correlated with training duration.
Koşaner et al., 2012 [35]	Preschool children with CI	Musical training	Prospective interventional study (pre-post)	Improved auditory–musical skills.
Welch et al., 2015 [36]	Children with CI/HA	Musical training	Pilot interventional study (single-group)	Improved pitch perception and singing skills.
Mason et al., 2021 [37]	Deaf children (mostly CI users)	Musical training	Interventional study (within-subject design)	Improved executive functions.

Table 3. Cont.

Study (Author, Year)	Population	Intervention Type	Study Design	Key Outcomes
Aksu et al., 2024 [38]	Preschool children with CI	Musical training	Prospective interventional study (pre-post)	Improved phonological awareness.
Dastgheib et al., 2013 [39]	Children with CI/HA	Musical training	Interventional study (pre-post)	Improved speech and language outcomes.
Xu et al., 2009 [40]	Prelingually deaf children with CI	Musical training	Controlled cross-sectional study	Significant deficits in vocal pitch accuracy; rhythmic production is comparable to normal-hearing controls.
Yucel et al., 2009 [41]	Children with CI	Musical training	Longitudinal interventional study (between-group control)	Improved musical perception (note discrimination).
Cason et al., 2015 [42]	Prelingually deaf children with CI	Musical training	Experimental study (within-subject design)	Rhythmic priming improved phonological accuracy (especially in CI users).
Rocca, 2015 [43]	Children with HL (pre- and post-CI)	Musical training	Observational descriptive study	Improved early communicative and listening behaviors.
Hidalgo et al., 2017 [44]	Children with HL (predominantly CI users)	Musical training	Experimental study (within-subject design)	Improved speech timing and auditory–linguistic processing.
Bedoin et al., 2018 [45]	Children with CI	Musical training	Controlled experimental study	Improved syntactic processing.
Lo et al., 2020 [46]	Children with CI/HA	Music therapy (multimodal program led by a certified therapist, integrating musical training)	Prospective controlled longitudinal study	Improved speech-in-noise perception, timbre perception, and prosodic processing; associated psychosocial benefits.
Lo et al., 2022 [47]	Children with CI/HA	Musical training	Prospective controlled longitudinal study	Reduced internalizing difficulties; improved parent-reported quality of life.
Gfeller et al., 2011 [48]	Preschool children with CI	Music therapy	Clinical descriptive study	Reported benefits in listening, speech, language, social, and emotional domains.
Esmailzadeh et al., 2013 [49]	Children with CI/HA	Music therapy	Interventional study (pre-post)	Improved language, cognition, and social interaction.
Barton & Robbins, 2015 [50]	Children with CI	Music therapy	Theoretical/clinical framework	Describes developmentally structured therapeutic techniques.
Comincini & Del Piccolo, 2013 [51]	Children with CI/HA; professionals and parents	Music therapy	Survey-based observational study	Perceived improvements in psychological well-being and engagement.

CI = Cochlear implant; HA = Hearing aid; HL = Hearing loss.

4.1. Music Therapy Approaches

Barton and Robbins began their work by outlining the potential of musical experiences to enhance developmental abilities in normally hearing children and subsequently described methods and music therapy approaches tailored to this population, with the aim of fostering auditory learning in early childhood [50].

Given the potential of music to positively influence brain regions involved in non-musical functions, such as attention, emotion, cognition, behavior, communication, and perception [52,53], two therapeutic frameworks for initiating auditory learning have been proposed: the Comprehensive Music Therapy Approach [54] and the Hybrid Approach to Pediatric Language Intervention [55]. This was followed by a presentation of specific musical and linguistic intervention techniques in which music was used as a supportive tool for working with children with CI.

Bruscia identified two main strategies that clinicians could employ within the Comprehensive Music Therapy Approach [54]: an “outcome-oriented strategy”, in which the therapist defines specific objectives based on the patient’s needs and applies evidence-based practices to address them, and an “experience-oriented strategy”, in which the therapist allows the patient to guide the process, using musical experiences as a means of revealing and shaping therapeutic goals. Specifically, four principal therapeutic methods are described [54]. The “re-creative method” involves the child singing or playing pre-existing songs, thereby supporting the development of specific skills and behaviors. The “improvisational method” allows the child to create melodies and rhythms spontaneously, offering an important channel for emotional expression, particularly in individuals with limited verbal abilities. The “receptive method” is based on active listening, with the child responding verbally or through movement, and can be used to stimulate imagination or to target specific auditory functions, such as associating movement with particular sounds or instruments. Finally, the “compositional method” involves the co-creation of musical material with the therapist, sometimes resulting in recordings or audiovisual products, and is particularly useful for expressing and documenting thoughts, feelings, and experiences.

Fey suggested that “natural” language interventions aligned with real-life communicative experiences increased the likelihood that skills would generalize to other contexts, supporting their long-term maintenance. Naturalness may be compromised in high-intensity, repetitive, exercise-based practices. Fey therefore proposed a “hybrid” approach to pediatric language intervention, balancing structured exercises aimed at acquiring basic musical skills with natural interactions in which the child actively “makes music,” similar to the Comprehensive Music Therapy Approach [55].

Barton and Robbins suggested a range of techniques in which music is used to support work with children with CI, organizing the interventions according to age group [50]. For infants and toddlers, they emphasize the use of infant-directed singing and songs integrated throughout the child’s daily routine, along with teaching nursery rhymes or songs associated with gestures, and highlighting the importance of moments of silence. For preschool children, the approach includes daily singing, the use of simple and accessible musical instruments, and the introduction of *sprechgesang*, a vocal technique that blends speech and singing. It also involves singing familiar melodies using different voices, incorporating musical games to reinforce listening skills, and using music to strengthen relational actions and conceptual understanding. With primary school children, the techniques shift toward transforming directive phrases into melodies or rhythmic patterns, working on synchronization with a steady beat, and using music as a tool to reinforce rhythm recognition. For upper primary school children, music is used more abstractly and expressively, such as conveying non-musical information through musical means, creating experiential opportunities for social interaction, encouraging musical com-

position to express feelings or personal experiences, and training skills like rhythm and frequency discrimination.

In the study by Gfeller and colleagues [48], possible music therapy interventions for preschool CI users were described, aimed at supporting listening skills, speech production, language development, socialization, cognition, emotional, behavioral, and motor development. Similar to the approaches outlined by Barton and Robbins [50], the authors emphasized adapting early-childhood musical activities to the specific needs of CI users and selecting “CI-friendly” musical stimuli. After describing empirical findings from studies on musical perception in CI users (covering rhythm, timbre, pitch, melody, song lyrics, singing, enjoyment, and musical participation), Gfeller and colleagues suggested collaboration between the music therapist and the broader clinical team [48]. This ensured that the therapist fully understood the child’s specific functioning while also providing assessments in previously unexamined areas, such as social skills. Consistent with Bruscia’s framework [54], key therapeutic targets included listening, speech production, and language development, addressed through the structured use of instruments, singing, and movement. The music therapist worked on key speech sounds by selecting specific instruments and encouraging the child to pronounce their names. Vocabulary and linguistic concepts were reinforced by using instruments to represent shapes, sizes, and physical characteristics, providing concrete examples. Interactive communication was also trained by asking the child questions about the instruments [48].

Movement-based activities were described as essential not only for motor development but also for supporting communication goals, echoing principles of naturalistic intervention proposed by Fey [55]. These activities could be directed toward sound detection, discrimination, identification, auditory comprehension, or language development. Musical stimuli effectively guided the speed and mode of movement, while the child’s movements could promote attention, perception, and understanding of a given stimulus. Because movement activities primarily supported listening skills, therapists were required to pay particular attention to providing clear verbal instructions at an appropriate pace, demonstrating new activities simply and clearly, or offering verbal instructions without background music. Lip-reading was also effective when the therapist’s face was visible. Singing, on the other hand, required coordination of the same vocal mechanisms used in spoken communication. Singing in group settings demanded attentive listening, which could be particularly challenging for children with HL. Songs were selected to reinforce vocabulary, speech sounds, and concepts relevant to the child’s daily life and individualized educational or therapeutic plan. Collaboration with audiologists or speech-language pathologists was recommended for setting objectives and selecting song texts, which could be adapted to meet specific needs [48].

Esmailzadeh and colleagues described the Auditory–Verbal Music Play Therapy (AVMPT), an educational approach based on listening and aimed at cognitive development [49]. This method integrated principles from Auditory–Verbal Therapy (AVT), play therapy, and music therapy. As originally articulated by Pollack in 1970 [56], AVT applies techniques, strategies, and procedures to promote spoken language development through listening, supporting children (even very young ones) in hearing, understanding, and communicating verbally. Parents are involved as primary facilitators of listening development through active and consistent participation in the child’s individual AVT sessions. According to the Association for Play Therapy (APT), play therapy is a structured, theory-based therapeutic approach in which trained clinicians use the inherent therapeutic value of play to help individuals address psychosocial difficulties and support healthy development. Esmailzadeh et al. reported that this type of intervention could benefit areas such as problem solving, behavior, mood, language, social skills, learning, and the expression of

feelings and fears. It was considered particularly suitable for children with HL, as it enabled them to express thoughts and emotions through the language of play in a context relying more on the visual channel than on spoken language [49]. In AVMPT, core principles of AVT, play therapy, and music therapy are integrated. Specifically, the music therapist teaches linguistic elements through musical games. Target words are introduced using strategies such as sing-song voice, modeling correct language, repetition, proximity to the microphone, repeating the word in various phrases, intoning or rhythmically producing it, and pairing verbal production with games or activities. Through these methods, the child may more effectively learn the target word and subsequently apply it in everyday communicative situations.

4.2. Summary of Evidence

Comincini and Del Piccolo conducted a two-part survey [51]. The first part involved administering a questionnaire to 60 professionals who treat children with HL participating in music therapy (including audiologists, speech therapists, otolaryngologists, general practitioners, and psychotherapists), aimed at understanding their opinions regarding this type of approach. The second part involved a group of 8 parents of children aged 3 to 6 years with varying degrees of HL who had attended weekly music therapy sessions for one year. Among professionals, 42% had not treated children with HL involved in music therapy and therefore did not express an opinion. Of the remaining participants, only a minority had direct clinical experience, but overall perceptions were positive. The main reported benefit was improved psychological well-being, whereas effects on sound discrimination, language, and prosody were considered less relevant. No resistance toward music therapy was observed among professionals.

In the parent survey, information was collected on participation characteristics (duration, modality, parental involvement) and perceived outcomes. Children were generally willing to participate in sessions, and parents reported predominantly positive emotional states following therapy, including happiness, calmness, and increased engagement. Overall, music therapy was perceived as effective in supporting auditory-behavioral rehabilitation and enhancing interaction with the environment.

Lo and colleagues investigated the benefits of a 12-week musical training program for children with HL, with a particular focus on psychosocial outcomes and quality of life [46]. Two groups of participants were tested: a group of children with sensorineural HL (SNHL) and a control group. The SNHL group undergoing the program consisted of 14 children (7 females and 7 males), aged between 6 and 9 years, with bilateral prelingual moderate-to-profound SNHL. Among them, 8 were bilateral CI users, 4 bimodal users, and 2 bilateral HA users. Eleven children started the musical training, while 3 completed only the baseline measurements. Of the 11 who began the training, only 9 completed all test sessions. The control group included 16 TH children (7 females and 9 males), aged 6–9 years. The program included weekly 40 min group music therapy sessions, with groups of 4–5 children led by a registered music therapist from the Speech and Hearing Clinic at Macquarie University. The approach followed was Nordoff-Robbins Creative Music Therapy, emphasizing interactive, social, and group-based activities in a non-verbal context. Additionally, 3 home sessions per week were prescribed, lasting 15–30 min, with online musical activities delivered via the MusicFirst Junior app, adaptable by the music therapist to meet specific objectives.

Assessments were conducted at baseline, after the 12-week training, and at a 12-week follow-up post-intervention. Results showed that, at baseline, children with HL had more emotional and behavioral difficulties and fewer prosocial behaviors than their TH peers. After the intervention, internalizing problems significantly decreased, and this improve-

ment was maintained at follow-up. Total difficulties also improved after the program, although this effect was not sustained over time. Importantly, after training, children with HL reached levels comparable to their TH peers in these areas. No significant changes were observed in externalizing problems or prosocial behavior. In terms of quality of life, children with HL initially scored slightly lower than their peers, and no significant improvements were observed following the intervention. Similarly, self-reported hearing-related quality of life remained lower than that of TH children, with no significant changes over time. However, parent-reported outcomes indicated that there were notable improvements in overall quality of life, particularly in emotional well-being, learning, confidence, and the ability to accomplish tasks, while no meaningful changes were found in physical health or vitality.

5. Limitations

This narrative review has several limitations that should be acknowledged. First, the available literature on music-based interventions in children with HL remains heterogeneous in terms of study design, sample size, participant characteristics, and outcome measures. Moreover, the number of available studies is still relatively limited, particularly for music therapy interventions in pediatric populations. Given this heterogeneity, a formal systematic synthesis or quantitative comparison of results would not have been methodologically appropriate. This variability limits direct comparison across studies and precludes firm conclusions regarding efficacy, particularly for music therapy interventions, which are still underrepresented in the literature. In contrast, evidence from adult populations is comparatively robust and well established [57–65]. Second, many of the reviewed studies involve small samples, short program periods, and a lack of long-term follow-up, which limits the ability to assess the durability and broader applicability of the observed effects and reduces the statistical power of the findings. Although control groups are frequently included, methodological aspects such as blinding procedures and effect size reporting are inconsistently described. Consequently, causal inferences regarding the impact of musical exposure remain limited. In addition, parental involvement, home practice adherence, and baseline musical exposure are often insufficiently controlled or reported, representing potential confounding factors. Third, outcome measures vary widely and frequently rely on parent- or self-reported questionnaires, which may be subject to reporting bias. Objective auditory or neurophysiological measures are less commonly employed, particularly in studies focusing on psychosocial outcomes. A further limitation is that audiological management (e.g., HA or CI fitting, optimization, and usage) was inconsistently reported across studies, making it difficult to determine whether the observed benefits of music-based interventions are influenced by appropriately fitted amplification or occur independently of it. The lack of studies comparing music-based family interventions with other family-centered approaches also limits the ability to disentangle the specific effects of music from those related to increased parent–child interaction and enriched listening environments. Finally, the inclusion of hybrid measures combining music therapy and musical training may have contributed to some overlap between categories, reflecting the current lack of standardized definitions in this field. These limitations highlight the need for future well-powered, longitudinal, and controlled studies using standardized outcome measures to better define the role of music-based interventions, and particularly music therapy, within pediatric hearing rehabilitation.

6. Clinical Perspectives: Development of an Early Music Therapy Program

Building on the evidence synthesized in this review, it is also important to consider how music-based approaches can be translated into structured clinical practice. Against this background, a pilot music therapy initiative was recently introduced at our pediatric audiology outpatient clinic within the Fondazione IRCCS Ca' Granda Ospedale Maggiore Policlinico in Milan (Italy). The project involves an early group music therapy intervention addressed to preschool-aged patients with CI or HA, starting from one year of age. Groups consist of a minimum of 3 to a maximum of 5 children, each accompanied by a parent who actively participates in the session. Sessions are held on a weekly basis, last 40 min, and are conceived within a medium- to long-term perspective (minimum duration of therapy: one year). The intervention is performed by a qualified professional music therapist, supported by a music therapist in training, and alternates structured activities of auditory–musical stimulation with moments of free instrumental and vocal improvisation. The approach is playful and multisensory, allowing ample space for expressiveness and interpersonal interaction. During the sessions, an observer external to the therapeutic setting (either a medical resident or a music therapist in training) is present and observes through a one-way mirror. Treatment monitoring is currently based on two instruments developed by the outpatient clinic team: an evaluation grid completed at the end of each session by the therapist together with the external observer, and a questionnaire periodically completed by parents and, separately, by the therapist. These tools aim to explore different domains, including engagement, relational and emotional aspects, as well as auditory, expressive, and communicative skills. Parental questionnaires are also intended to explore perceived changes across several developmental areas, including motor, linguistic, cognitive, relational, emotional, sound-musical, and expressive-creative domains.

This clinical initiative is currently being implemented as part of the broader rehabilitative pathway offered to children with HL using CI or HA. As the program expands, systematic longitudinal monitoring and structured outcome analyses will be essential to delineate its specific contributions to auditory, communicative, emotional, and relational development. In a broader perspective, initiatives of this kind may help shape future evidence-informed frameworks for integrating music therapy into multidisciplinary pediatric hearing rehabilitation, ultimately contributing to the definition of standardized, developmentally sensitive clinical protocols.

7. Conclusions

Drawing on studies concerning musical auditory perception in children with HL, it is evident that music plays an important role as a primary perceptual stimulus, closely linked to the early development of auditory and language functions, as well as to cognitive and emotional growth. Limitations in musical perception among children with HL, particularly CI users, are well documented and primarily affect timbre and pitch perception, whereas rhythmic abilities appear relatively preserved. However, engagement in activities aimed at enhancing musicality has been reported to improve musical perception, leading to greater enjoyment of music and, consequently, enhanced overall quality of life. Music-based interventions have also been associated with improved auditory perception and verbal skills. Therefore, identifying adequate and effective musical intervention strategies that engage pediatric patients early and positively, and that allow for medium- to long-term treatment durations, is crucial. In this context, musical training interventions are far more prevalent than music therapy interventions. Notably, studies on musical training predominantly involve school-aged children, while research on preschoolers is very limited.

Consequently, parental involvement is minimal, reported in only one study, where parents' role was limited to supervising online musical exercises at home. Within the musical training domain, the presence of control groups is significantly lower than in studies on musical auditory perception. In some cases, treatment efficacy is assessed via pre- and post-intervention tests administered to the children or their parents. Identifying control groups with homogeneous characteristics (e.g., age or clinical history) is often challenging, which may explain the frequent use of within-subject control designs. From a musical perspective, key outcomes include improvements in timbre and pitch perception (the latter positively correlated with the duration of musical education) and note discrimination abilities. Positive effects on linguistic and syntactic skills appear more evident in training programs focused on rhythm.

Regarding music therapy, the literature is sparse, with a limited number of studies available. Among these, only one reports a clinical trial, which employed an integrated approach combining group music therapy and musical training. This was a short-term intervention, partially compatible with music therapy principles, which typically require medium- to long-term application. Consequently, the efficacy of music therapy in pediatric populations with HL remains insufficiently documented. This may reflect limited awareness among healthcare professionals of the potential benefits of music therapy in children—not only for auditory perception but also for attention, emotion, cognition, behavior, and communication. Such benefits appear closely linked to the naturalness and enjoyment of the activity, which integrates auditory perception with other perceptual channels and provides ample scope for movement, play, and interpersonal interaction. Although limited in number, parent perceptions of children undergoing music therapy are qualitatively positive, encompassing auditory, behavioral, and social aspects. The sporadic availability of studies on music therapy effectiveness highlights the need for increased experimental music therapy projects within healthcare facilities serving children with HL, involving team-based planning and outcome assessment. Finally, careful consideration of evaluation methods is warranted, given the wide variety of pre-existing and ad hoc scales and questionnaires used in the reviewed studies. Notably, assessments in musical training interventions focus primarily on auditory perception, musical parameters, and language, whereas those in music therapy also consider broader aspects of quality of life. The use of standardized assessment scales, while useful for objective and widely comparable evaluation, should be complemented by questionnaires and rating grids capturing all dimensions of music therapy interventions, including socialization, behavior, emotional regulation, and overall patient well-being.

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Article

Endoscopic Myringoplasty for Pediatric Tympanic Membrane Perforations: Is It Worth It?

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Abstract: Background/Objectives: The endoscopic repair of the tympanic membrane is an established method for addressing perforations in the tympanic membrane. However, there are limited studies in the literature examining the success rate of this procedure in the pediatric population. **Methods:** This study retrospectively analyzed data from the tertiary referral center at the University Hospital of Verona, Italy. This medical record contains data from 84 type 1 transcanal endoscopic tympanoplasties performed in pediatric patients between November 2014 and February 2022. Seventy-seven pediatric patients aged 4 to 16 years who underwent type 1 transcanal endoscopic tympanoplasty (seven of whom underwent bilateral surgery at different time points) were included in the study. Our study did not include more extensive procedures than type 1 endoscopic tympanoplasty. Only patients with tympanic membrane perforation due to simple chronic otitis media, trauma or when no apparent cause was found were included. Chronic otitis with cholesteatoma and other pathologies of the external or middle ear were exclusion criteria. Patients with a follow-up of less than 12 months were excluded from this study. The technique was based on the endoscopic placement of an underlay graft of temporal fascia or tragal cartilage to repair a tympanic membrane perforation. Demographic, clinical, audiologic, and surgical data were collected from each patient. In the study, we considered the reduction of the air-bone gap (ABG) as a functional outcome and the integrity of the reconstruction as an anatomic outcome of success. **Results:** The primary surgery had a closure rate of 92.9% (78 of 84). All patients underwent audiological evaluation 4–6 months post-surgery, with 84 ears tested. The mean preoperative ABG was 17.13 dB HL, reduced to 9.16 dB HL postoperatively, showing a mean reduction of 7.97 dB HL. No significant complications occurred. **Conclusions:** Transcanal endoscopic type 1 tympanoplasty should be considered a safe procedure with a high success rate for the repair of tympanic membrane perforations, even in pediatric patients.

Keywords: myringoplasty; tympanoplasty; tympanic membrane perforation

1. Introduction

Since the introduction of ear surgery, tympanic membrane repair has traditionally been achieved by microscopically assisted surgery [1]. In the last decade, the endoscope has become an alternative tool in treating otologic pathologies, and endoscopic ear surgery has taken its place as one of the techniques of choice in treating middle ear cholesteatoma. Currently, type I endoscopic tympanoplasty is widely performed worldwide because of the many advantages of this technique. Perforations of the tympanic membrane are common

problems ENT physicians encounter in their practice and are among the most common surgical procedures performed worldwide. This procedure has two main goals:

- full closure of tympanic membrane perforations;
- enhancing hearing level and reducing the air-bone gap (ABG).

In recent years, several studies have shown that the endoscopic technique is as effective or even more effective than the microscopic technique in closing tympanic membrane perforations. Marchioni et al., in a previous article [1], demonstrated an 86% closure rate in a case series of 98 patients, with improvement in ABG within 20 dB HL in 89% of patients. However, most of these studies were performed in adults [1–14]. There is a lack of studies in the literature examining the effectiveness of this procedure in children. One of the most important studies we found was by Kaya et al. [15], in which type I tympanoplasty was performed on both ears with different techniques (microscopic on one side and endoscopic on the other), and the endoscopic technique showed similar results and even better results in terms of pain management and operation time. Many studies have shown controversial results regarding age. Friedberg et al. [16] suggest that the proper age range for performing tympanoplasty is between 7 and 11 years, as they did not record higher graft failure rates in this range. These results may be due to the complete maturation of the eustachian tube at this age. This study describes the endoscopic procedure used at our institution to treat tympanic membrane perforations in children, regardless of size or location. The present study is currently one of the most comprehensive published case series in the pediatric population because the technique was introduced relatively recently. Additional studies with a larger patient cohort are required to validate the findings presented in this paper.

2. Materials and Methods

A retrospective cohort study was conducted on 77 pediatric patients, aged 4 to 16 years, who underwent type I endoscopic tympanoplasty between November 2014 and February 2022. This study took place in the Department of Otorhinolaryngology and Head and Neck Surgery at the University Hospital of Verona. The following standardized surgical procedure was employed to treat consecutive patients with tympanic membrane perforation, and it was executed by surgeons in the ENT Department at the University Hospital of Verona. Patients who required a more extensive procedure than type I endoscopic tympanoplasty were not included in our study. The study included only patients diagnosed with chronic otitis media with tympanic membrane perforation, post-traumatic tympanic membrane perforation, or idiopathic perforations. Those who were found to have cholesteatoma preoperatively or intraoperatively were excluded. All patients had perforations involving more than one quadrant. This study did not include patients with minor perforations who underwent fat graft myringoplasty. The follow-up period was at least 12 months; patients whose follow-up time was shorter were excluded from the study. Each patient underwent an endoscopic examination of the ear before surgery. Data on perforation location and size were categorized into anterior, posterior, inferior, and subtotal areas. The data analyzed regarding the surgery included graft types (tragus or concha cartilage, temporalis fascia, bovine pericardium collagen acellular matrices), intraoperative ossicular chain abnormalities, and complications (intraoperative and postoperative). Subsequent evaluations involved endoscopic examinations of the external auditory canal and tympanic membrane at one month and six months post-surgery, with data gathered on tympanic membrane healing, infection rates, reperforation occurrences, and the need for revision surgeries. Audiometric evaluations were conducted to assess hearing function before surgery and 4 to 6 months postoperatively. Hearing outcomes were measured by comparing pre and postoperative air and bone pure tone averages (PTA) at frequencies of 0.5, 1, 2, and 3 kHz. For each patient, the PTA score and air-bone gap (ABG) were calculated both pre and

postoperatively. All procedures were carried out by three experienced surgeons, each with a minimum of 8 years of expertise in endoscopic ear surgery.

2.1. Preoperative Considerations

Prior to planning an endoscopic transcanal ear exclusive approach, it is essential to consider the patient's age, the condition of the perforation and tympanic cavity, as well as any previous surgeries for tympanic membrane perforation.

2.1.1. Age

Although age is not an absolute contraindication for endoscopic ear surgery, special caution should be exercised when performing type I endoscopic tympanoplasty on children under six years of age. This precaution is necessary due to the incomplete maturation of the rhinotympanic system and the relatively small size of the external auditory canal (EAC). Children have a higher likelihood of developing infections that can lead to a new perforation and potential failure of the surgery. Nevertheless, the straight shape of the ear canal in children provides a favorable condition for performing endoscopic ear surgery.

2.1.2. Perforation and Condition of the Tympanic Cavity

Type I endoscopic tympanoplasty was not conducted if unstable conditions were present in the tympanic cavity, such as infection or recurrent otorrhea, because these conditions could adversely affect the closure rate.

2.1.3. Graft Selection

Suitable grafting materials include cartilage, temporalis fascia, vein grafts, fascia lata, and perichondrium. Temporalis fascia is the most commonly used and has a high success rate for closures. Cartilage grafts have recently been increasingly used, especially in complex perforations or revision procedures. In our study, the graft material was selected similarly to what we chose for adults (as shown in Table 1), using cartilage grafts for complex cases, such as anterior or more than two quadrant perforations [17]. Xenografts are tissues from non-human species that have been engineered for use as graft material in type I tympanoplasty, intended for tympanic membrane repair similarly to temporalis fascia. Acellular collagen matrices derived from bovine pericardium (Tutopatch—Tutogen Medical Inc., Alachua, FL, USA) are utilized by our department.

Table 1. Choice of graft material.

Graft Material	Indications
Temporalis fascia	Perforation confined to one sector of the tympanic membrane (better for posterior perforations)
Tragal cartilage	Wide perforations with more than one affected sector or involvement of the annulus or failure of the fascia temporalis
Conchal cartilage	Earlier failures when no tragal cartilage is available
Xenograft	Never used as first choice, sometimes used to reinforce reconstruction

2.1.4. Compliance with Ethical Standards

This study complied with the ethical standards of the institutional and national research committees, as well as the 1964 Declaration of Helsinki and its amendments. Informed consent was obtained from all participants.

2.1.5. Surgical Procedure

Position of the Patient

The patient's positioning remains consistent with that employed for microscopic surgery. The skin of the auricular region and external auditory canal is prepared using a povidone-iodine solution. The primary surgeon utilizes the endoscope with their non-dominant hand while manipulating surgical instruments or suction with their dominant hand. The endoscopes utilized included a 0°, 3-mm-width, 14-cm-length endoscope (Karl Storz, Tuttlingen, Germany) or a 0°, 4-mm-width, 18-cm-length endoscope. In cases where middle ear pathology, particularly middle ear cholesteatoma, was suspected, a rigid 45° endoscope (45°, 14 cm, 3 or 4 mm wide) was used to examine the anatomy of the middle ear and concealed areas in greater detail. The tympanomeatal flap design is tailored to the defect's location and size.

Surgical Steps

Most of the procedures were performed utilizing a 0°, 3-mm, 14-cm endoscope. Initially, the external auditory canal (EAC) is infiltrated with a solution of 2% mepivacaine and epinephrine, administered 3 min before the procedure. To facilitate graft integration, scarring of the perforation margins is performed using an otologic sickle knife. An incision is created using an otologic round knife in the external auditory canal (EAC) skin, approximately 5–10 mm from the annulus, positioned between the 6 and 12 o'clock marks. The primary incision can be initiated with a monopolar scalpel to create a dotted line for coagulation and reduced bleeding, and then completed with a round knife, based on the surgeon's preference. The tympanomeatal flap is carefully detached from the EAC bone and elevated until the annulus is visible, using the round knife and a cotton ball soaked in epinephrine to protect the tympanic membrane and control bleeding. Any bleeding is promptly managed with epinephrine-soaked cotton balls applied for one minute. The mucosa of the middle ear is then incised, and the fibrous annulus is elevated using a cotton ball while preserving the integrity of the chorda tympani. The flap is lifted from posterior to anterior, remaining attached to the malleus handle. Complete visualization of the middle ear cavity is achieved after detaching the posterior malleolar ligament. In the case of posterior perforations, it may not be required to completely detach the tympanic membrane from the malleus. Adequate management of the anterior margin of this type of perforation can still be achieved. In cases of anterior perforations, the periosteum is detached from the handle of the malleus using a sickle knife. The flap is then separated from the ossicular chain at the subperiosteal level while leaving the umbo attached. Micro-scissors are used to separate at the umbo. The membrane is moved forward while staying connected to the anterosuperior wall of the external auditory canal. The mobility of the ossicular chain is then assessed. A thorough examination of the entire tympanic cavity is conducted using angled endoscopes to rule out middle ear pathology (e.g., cholesteatoma) and detect possible isthmus obstruction, which can affect middle ear ventilation and subsequent procedural success. The chosen graft is carefully shaped to match the size of the perforation and is then inserted laterally on the malleus handle using a sickle knife, ensuring that the edges remain beneath the fibrous annulus (underlay technique). In instances of posterior perforation, the graft can be positioned medial to the malleus handle without totally detaching the flap. When utilizing temporalis fascia as the graft material, it is crucial to compress and adequately dry it to enhance its stiffness and maneuverability, which is particularly essential for sliding the graft under the anterior segment of the tympanic tube and the fibrous ring. For cartilage grafts, the perichondrium is left intact on one side, with approximately 1 mm overhanging the posterior edge of the cartilage and removed from the other side. The extracted perichondrium is preserved in case another graft becomes necessary, with

the perichondrium being pressed and dried similar to the fascia. Once the graft is precisely shaped, an incision is crafted to accommodate the malleus handle and the posterior sulcus. The cartilage is then inserted, ensuring that the perichondrium side contacts the tympanomeatal flap, and the perichondrium of the posterior edge of the cartilage graft aligns with the posterior wall of the external auditory canal (EAC). If temporal fascia is used, the graft is elevated anterosuperiorly and secured to the tympanomeatal flap. Gel foam is introduced into the tympanic cavity to serve as a scaffold for the graft, with care taken to prevent obstruction of the eustachian tube or isthmus by scar tissue. The graft and tympanomeatal flap are meticulously repositioned. The endoscopic view provides excellent visualization of the anterior edges of the annulus fibrosus ring and its position relative to the graft, ensuring optimal outcomes. Small quadrangular pieces of Gelfoam are positioned at the edges of the perforation and flap incision to assist in healing, protect the graft, and keep it in place. Subsequently, the external ear canal is filled with Gelfoam pieces, ensuring no pressure is applied to the tympanic membrane.

2.1.6. Postoperative Care and Follow-Up

If no complications occur, patients are discharged the next day. They should avoid actions that increase middle ear pressure, such as Valsalva maneuvers, sneezing, abdominal contractions, and nose blowing. Intense exercise and high-altitude flying are discouraged. Patients are advised to avoid getting water in the operated ear and to apply antibiotic ear drops twice daily for a month to prevent infections. A follow-up endo-otoscopic exam is conducted one month post-surgery to confirm healing before resuming normal activities. Audiometry tests are performed four months after surgery to assess results. Annual endo-otoscopy is recommended for at least two years. After discharge, it is crucial for patients to adhere strictly to post-operative instructions to ensure optimal healing and to prevent complications. Any signs of infection, such as fever, increased pain, or unusual discharge from the ear, should be reported to the healthcare provider immediately. While using antibiotic ear drops, ensure the medicine reaches deep into the ear canal by tilting the head sideways for a few minutes after application. For those who experience discomfort or mild pain, over-the-counter pain medications like acetaminophen or ibuprofen may be used unless contraindicated. It is also advisable to avoid environments with extreme temperatures or humidity, which could potentially affect the ear's healing process.

3. Results

Seventy-seven pediatric patients were included in our study, for a total of 84 procedures (seven bilateral procedures) completed between November 2014 and February 2022 at the Department of Otolaryngology, Head and Neck Surgery of the University Hospital of Verona. Of the 77 patients selected, 12 had a bilateral perforation, but only 7 underwent bilateral surgery, bringing the number of type I tympanoplasty procedures to 84. Regarding laterality, 42 right (50%) and 42 (50%) left perforations were treated. The male-to-female ratio was 49:35. The mean age at surgery was 10.2 years (range 4–16 years). Perforations were categorized according to size and location and were divided into the following categories: anterior (30 perforations, 35.7%), inferior (23 perforations, 27.4%), posterior (9 perforations, 10.7%) and subtotal (22 perforations, 26.2%). In all patients, type I endoscopic tympanoplasty was performed using only the abovementioned technique. The graft material chosen was mainly autologous, with tragal cartilage in 34 cases (40.5%) and autologous temporalis muscle fascia in 43 cases (51.2%) (Table 2). In all cases where only tragal cartilage was utilized, no additional perichondrium was necessary, as the provided cartilage was sufficient. In our study, two complications were observed, one intraoperatively (chorda tympani section) and one postoperatively (iatrogenic cholesteatoma limited

to mesotympanum which needed a revision surgery through an endoscopic exclusive transcanal approach). Endoscopic examinations were performed in all 77 patients (84 surgeries in total) 1, 6 and 12 months after surgery. Persistent perforation after surgery occurred in only six (7.14%) patients 1 month after surgery. Among the failures, one occurred in a patient with preoperative subtotal perforation (1 out of 22, or 4.5%), and five occurred in patients with preoperative anterior perforation (5 out of 30, or 16.67%). Three failures were observed in patients with chronic otitis perforations, two failures in cases of idiopathic perforation, and one failure resulted from trauma. Thus, the closure rate of primary surgery was 92.85% (78 of 84). In all failed operations, the graft material of choice was temporalis muscle fascia. The average duration of the surgical procedure (the time from injection of local anesthesia to closure of the ear canal) was 69.5 (34–132) minutes for all 84 procedures in which endoscopic transcanal tympanoplasty type I was performed.

Table 2. Main results.

	n	%
TM Perforations	84	100.0
Side		
Left	42	50.0
Right	42	50.0
Gender		0.0
Female	35	41.7
Male	49	58.3
Mean age (years)	10.2	(4–16)
Perforation location		
Anterior	30	35.7
Posterior	9	10.7
Inferior	23	27.4
Subtotal	22	26.2
Graft		
Temporalis muscle fascia	34	40.5
Tragal cartilage	43	51.2
Temporalis muscle fascia + xenograft	3	3.6
Tragal cartilage + xenograft	2	2.4
Temporalis muscle fascia + tragal cartilage	1	1.2
Xenograft	1	1.2
Complications		
None	82	97.6
Chorda tympani damaged	1	1.2
Cholesteatoma	1	1.2
Surgery duration (minutes)	69.5	(34–132)
TM repair		
Yes	78	92.9
No	6	7.1
Initial perforation location in failure		
Inferior	5	6.0
Subtotal	1	1.1
Graft used in failure		
Temporalis muscle fascia	5	6.0
Temporalis muscle fascia + xenograft	1	1.1
BC PTA (mean)		range
Preoperative	9.24	(5–26.25)
Postoperative	8.61	(5–26.25)

Table 2. Cont.

	n	%
AC PTA (mean)		
Preoperative	26.69	(10–48.75)
Postoperative	17.77	(10–37.5)
ABG (mean)		
Preoperative	17.13	(5–33.75)
Postoperative	9.16	(10–26.75)
Ears with normal hearing (AC PTA < 20 dB HL)		
Preoperative	29 of 84	34.5
Postoperative	62 of 84	73.8
Pre vs post ABG		
>10 improvement	26	31.0
0–10 improvement	51	60.7
no change	4	4.8
0–10 worsening	3	3.6
>10 worsening	0	0.0

All patients were audiometrically assessed between 4 and 6 months postoperatively, and a total of 84 ears were tested. The mean preoperative ABG value was 17.13 dB HL, whereas the mean postoperative ABG value was 9.16 dB HL, corresponding to a mean ABG reduction of 7.97 dB HL. An ABG value of less than 20 dB HL was considered an appropriate audiological outcome. This was achieved in 91.7% (77 of 84) of cases, with 31% (26 of 84) of cases displaying a hearing improvement of at least 10 dB HL (in terms of ABG closure). Hearing performance worsened slightly in only 3.6% (3 of 84) of cases; no worsening > 10 dB HL was noted. It is important to note that there was no deterioration of bone conduction, which means that the interventions caused no sensorineural hearing loss. There were no changes in ABGs in 4.8% (4 of 84) of cases.

4. Discussion

Minimally invasive surgery is being adopted more frequently in various surgical disciplines. In the field of ENT surgery, the endoscope has been effectively integrated into otologic approaches. Traditionally, middle ear surgeries performed under microscopic vision are now frequently conducted using endoscopic techniques. The microscopic approach allows the surgeon to operate with both hands, which aids in tissue manipulation and bleeding control. On the other hand, the microscopic transcanal approach does not allow for complete exposure of the tympanic membrane and its medial surface, hindering visualization of the anterior margin of perforations. Conversely, the endoscope offers precise visualization of these areas. It offers a clear view of the tympanic membrane, especially in patients with intricate external ear anatomy, such as narrow and curved ear canals. The enhanced visibility afforded by the endoscope enables reliable refreshment of the perforation margins and successful grafting [18]. Furthermore, this technique typically obviates the need for canaloplasty even in patients with narrow or tortuous ear canals. It also reduces postoperative bleeding and pain, and yields superior cosmetic results compared to the traditional microscopic retro auricular approach, while minimizing local and general postoperative side effects. The transcanal endoscopic technique does not require cutting or damaging the vascular strip that supplies blood from the malleolar and radial arteries to the tympanic membrane, thereby facilitating faster wound healing [19]. Since 2014, our center has refined the endoscopic type I tympanoplasty technique for rapid flap formation, good bleeding control, and easy pad grafting. This study shows the feasibility and benefits of the transcanal endoscopic method in children. The technique successfully

repaired the tympanic membrane in all patients by treating the perforation margins. It can be performed regardless of perforation size or location, even with narrow or tortuous ear canals. In this series, no procedures required switching to the traditional microscopic technique. Among 77 patients and 84 procedures, the primary surgery closure rate was 92.85%. The literature reports overall closure rates between 84% and 96%, consistent with our findings [4,20,21]. The success rate for closure in type I tympanoplasty using microscopic techniques has historically ranged between 78% and 90% [21,22]. Improving hearing is crucial for enhancing future quality of life. Numerous studies have documented positive outcomes in postoperative hearing gain for patients who have undergone endoscopic type I tympanoplasty. Lade et al. [17] reported a mean ABG closure in pediatric patients undergoing type I endoscopic tympanoplasty of 10.37 dB versus a mean ABG closure of 15.5 with the microscopic technique, without statistical significance. Our results were similar to those observed in the literature. The mean preoperative ABG value was 17.13 dB HL, whereas the mean postoperative ABG value was 9.16 dB HL, corresponding to a mean ABG reduction of 7.97 dB HL. An ABG value of less than 20 dB HL was considered an appropriate audiological outcome. This was achieved in 91.7% (77 of 84) of cases. Hearing performance worsened slightly in only 3.6% (3 of 84) of cases; no worsening > 10 dB HL was observed. This means that type 1 endoscopic tympanoplasty has a very high success rate in closing tympanic membrane perforations and guarantees an excellent audiological outcome. A key benefit of endoscopic procedures in middle ear surgery is the reduction in surgical duration, which is significant for minimizing anesthesia time. This advantage may be attributed to the fact that endoscopic techniques do not necessitate sutures and allow for faster visualization of concealed areas. In our study, the mean duration of the surgical procedure was 69.5 min, with a range of 30 to 145 min. Some researchers suggest that the lateral placement of the fascia or cartilage graft on the malleus handle may pose a risk for delayed blunting of the neo-tympanum [23]. The over-under technique described by Kartush et al. [24], where the graft is positioned lateral to the malleus handle and medial to the tympanic ring, has shown effective functional results and closure rates in type I microscopic tympanoplasty. In this study, no cases of blunting were noted at follow-up, indicating the practicality of this technique. It highlights that precise placement of the graft under the annulus in its anterior region is crucial to prevent lateral displacement. Though the number of cases is too small for statistical analysis, we observe that five out of six surgery failures with reperforation were anterior, using temporalis muscle fascia as the graft. This may suggest that temporalis muscle fascia is not preferred when the perforation is anterior in children. Anterior perforations present a greater challenge to manage using both endoscopic and microscopic approaches. This may be considered a limitation of the technique. However, it is important to note that anterior perforations treated with cartilage have been successfully managed. Further research is required to validate these findings. This could be a valid topic of debate, especially considering that new techniques are emerging, such as tympanic membrane regeneration therapy used to treat tympanic membrane perforation (TMP) based on a tissue engineering method developed by Kanemaru et al. [25] In this treatment, the perforated TM is mechanically freshly wounded through the external auditory canal, and a gelatin sponge impregnated with trafermin (recombinant human basic fibroblast growth factor) is placed inside and outside the TM so that it contacts the perforated TM. This minimally invasive technique requires no skin incision or autologous tissue collection.

The present study is currently one of the most comprehensive published case series in the pediatric population because the technique was introduced relatively recently. Further studies with more patients are needed to confirm the data presented in this paper.

This study is retrospective and, therefore, has some limitations. One of the most important limitations is that there might be a bias in the selection of patients because the choice of the graft was based on our experience and did not follow the rules of randomization. Another limitation is the lack of microscopic control in type I tympanoplasty, even though microscopes are widely used and effective for other procedures such as stapedoplasty [26]. Thus, we had to rely on the general literature, which is often variable. We tried to exclude any obvious bias because this was a consecutive series of patients with strict inclusion and exclusion criteria, treated in the same way and with the same technique by three experienced surgeons at the exact center. In any case, this is one of the studies in the literature with the highest number of pediatric patients treated with EES for repair of MT.

5. Conclusions

Endoscopic ear surgery has now become a viable alternative to traditional microscopic ear surgery in many cases. Our center has one of the largest caseloads globally. While this study does not claim that the endoscopic technique is superior to the microscopic one, it demonstrates that type I transcanal endoscopic tympanoplasty can be considered a safe and effective technique with a high success rate for treating tympanic membrane perforations, even in young patients.

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Institutional Review Board Statement: The study was conducted in accordance with the Declaration of Helsinki. Approval from our Ethics Committee was not required, as this procedure is a well-known and accepted standard for treating tympanic membrane perforations, even in children.

Informed Consent Statement: Informed consent was obtained from all subjects involved in the study. In this case, considering that patients were minors (under 18 years old), consent was obtained from both mother and father. Written informed consent has been obtained from the patient(s) to publish this paper.

Data Availability Statement: The original contributions presented in this study are included in the article. Further inquiries can be directed to the corresponding author.

Conflicts of Interest: The authors declare no conflicts of interest.

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Article

Paediatric Utilisation of Ophthalmic Antibiotics in the Ear in Aotearoa/New Zealand

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Abstract: Background/Objectives: Some ophthalmic antibiotics are publicly subsidised in New Zealand (NZ) for off-label use in the ear, however, this utilisation has not previously been described. This study compared the utilisation of ophthalmic chloramphenicol and ciprofloxacin in the eye and ear, among NZ children. Methods: This study involved clinical record review, and included 11,617 prescriptions of ophthalmic chloramphenicol and ciprofloxacin in 2022, for children aged five years or under in Auckland, NZ. Prescriptions of chloramphenicol and ciprofloxacin for eye and ear use were compared by: patient age, gender, ethnicity and socioeconomic deprivation, indication, community or hospital prescribing and number of repeat prescriptions. Statistical analysis was performed using Chi-squared test and multinomial regression. Results: Most ophthalmic ciprofloxacin was used in the ear (84%). In contrast, almost all chloramphenicol was used in the eye (96%). Post-operative use following tympanostomy tube insertion accounted for half of all hospital-prescribed ophthalmic ciprofloxacin used in the ear. Utilisation of chloramphenicol and ciprofloxacin in the eye and ear was similar, with more prescriptions for children aged one year and males, and most children received only one prescription. Māori and Pacific children generally received fewer prescriptions. Pacific children were more likely than Māori children to receive hospital-prescribed ophthalmic ciprofloxacin for use in the ear (adjusted OR 6.7, $p = 0.025$). Conclusions: These findings highlight the utilisation of ophthalmic ciprofloxacin in the ear in NZ children. These findings will inform decision-making in the public funding of medications, policy development in equitable medication access, and more collaborative efforts to improve antimicrobial use.

Keywords: child; New Zealand; anti-bacterial agents; eye infections; otitis

1. Introduction

Paediatric utilisation of ophthalmic antimicrobials is relatively high [1], and these antibiotics are often used in the management of acute bacterial conjunctivitis [2], which is highly prevalent in the paediatric population. However, serious complications are rare [2], and most cases are self-limiting [3]. Antimicrobial therapy aims to speed resolution and decrease transmission; however, evidence supporting such use is limited [3].

Antibiotic exposure drives the development of resistance, and resistant organisms have been isolated from bacterial eye swabs from children [4]. Additionally, topical antibiotic use can alter the ocular surface microbiome in children [5], and changes to the microbiome have been found in some ocular surface conditions [6].

In New Zealand (NZ), chloramphenicol and ciprofloxacin are among the most commonly used ophthalmic antibiotics [7]. Of note, these ophthalmic formulations are also

publicly subsidised in NZ for off-label use in the ear. Topical antibiotics are often used in the ear in the management of conditions such as chronic suppurative otitis media [8], which is prevalent among children. However, there are concerns around the development of antimicrobial resistance and potential ototoxicity, related to antibiotic exposure in the ear [9,10].

There is limited evidence on the utilisation of ophthalmic antibiotics in the ear in NZ. As such, this study aimed to compare the use of ophthalmic chloramphenicol and ciprofloxacin between eye and ear conditions, with a focus on usage among young children. This study found the utilisation of ophthalmic ciprofloxacin in the ear to be common among NZ children.

2. Materials and Methods

Ethical approval for this study was obtained from the Auckland Health Research Ethics Committee (reference AH24592). Institutional approval was obtained from the Te Whatu Ora Auckland Research Review Committee (reference A+9733). As this cross-sectional study involved retrospective analysis of hospital records only, and did not involve working with human participants, informed consent and Declaration of Helsinki are not applicable for this analysis.

This study included every prescription of 0.5% chloramphenicol and 0.3% ciprofloxacin eye drops between January and December 2022, for children aged five years or under with a public hospital record, within Health NZ-Te Whatu Ora Auckland, Counties Manukau and Waitemata health districts in Auckland, NZ. This analysis included medications prescribed and dispensed in the community, and medications prescribed during a hospital visit for subsequent dispensing in the community. In-hospital medication use and hospital-supplied medications were excluded from this analysis.

The proportion of chloramphenicol and ciprofloxacin prescriptions for use in the eye and ear was quantified; site of use was ascertained from the prescribing notes. The proportion of community and hospital prescriptions was also compared between eye and ear conditions. Specific indications for chloramphenicol and ciprofloxacin use were ascertained for hospital prescriptions; as this study analysed hospital records, indications for community prescriptions were unavailable and unable to be included in this analysis.

Number of prescriptions within the study period and patient age, gender, ethnicity and socioeconomic deprivation were compared between eye and ear use for chloramphenicol and ciprofloxacin. Proportion of patients receiving one or multiple prescriptions was compared. Proportion of prescriptions by age in years, gender, ethnicity and deprivation quintile was compared. Neighborhood deprivation was based on NZDep2013 Index of Deprivation decile [11], and deciles were aggregated into equally sized quintiles. Population proportions by gender and ethnicity were obtained from the 2018 Census.

The following factors were quantified based on every prescription included in this study: proportion of prescriptions for eye and ear use, number of prescriptions within study period and patient age. Due to the large number of prescriptions and manual extraction of the following data, the following factors were quantified based on a subset of 300 randomly sampled prescriptions, for chloramphenicol for eye use and ciprofloxacin for ear use: proportion of hospital and community prescriptions, specific indication and patient gender, ethnicity and deprivation. As there were fewer than 300 prescriptions of ciprofloxacin specified for eye use, and of chloramphenicol for ear use, these factors were quantified based on every prescription included in this study.

Data and statistical analyses were performed using IBM SPSS Statistics version 29 (Armonk, NY, USA). Missing data was omitted from analysis. Chi-squared test was performed to determine significant differences in proportions of prescriptions between

comparison groups. Multinomial regression was performed to determine independent and significant associations between: (1) receiving a hospital prescription and age, gender, ethnicity and deprivation; (2) receiving a prescription before one year of age and gender, ethnicity and deprivation; and (3) receiving multiple prescriptions and gender, ethnicity and deprivation. As such, potential confounders such as ethnicity and deprivation were adjusted for in this regression analysis. $p \leq 0.05$ was considered to be statistically significant.

3. Results

This analysis captured 10,227 chloramphenicol and 1390 ciprofloxacin prescriptions. Notably, most ciprofloxacin was used in the ear (84%), and only 9% was for ophthalmic use ($n = 1168$ and 125 respectively). In contrast, almost all chloramphenicol was for ophthalmic use (96%), and only 0.3% was used in the ear ($n = 9818$ and 31 respectively). Site of use was not specified for 3.7% ($n = 378$) and 7% ($n = 97$) of chloramphenicol and ciprofloxacin prescriptions respectively, and these were not analysed further.

The following factors were quantified based on every prescription with specified site of use, as described above: number of repeat prescriptions and patient age. Due to the large number of prescriptions of ciprofloxacin for ear use and chloramphenicol for eye use, other factors were quantified based on subsets of 300 randomly selected prescriptions: proportion of hospital and community prescriptions, specific indication and patient gender, ethnicity and deprivation. For ciprofloxacin for eye use and chloramphenicol for ear use, quantification of these factors was based on every prescription as there were fewer than 300.

Most antibiotics were community-prescribed (Table 1); however, the proportion of hospital prescriptions was significantly larger for usage in the ear, compared to ophthalmic use ($p < 0.001$). Almost all chloramphenicol for ophthalmic use was community prescribed (98.7%).

For hospital prescriptions, a large proportion of ciprofloxacin used in the ear (50%) was attributed to post-operative use following tympanostomy tube insertion (Table 2). Otitis externa, otitis media and otorrhea also contributed to ciprofloxacin and chloramphenicol usage in the ear. Indications for ophthalmic use of ciprofloxacin included corneal ulcers, microbial keratitis and bacterial conjunctivitis, while indications for ophthalmic use of chloramphenicol included viral conjunctivitis.

Most children received only one prescription over the study period (Table 1). However, with ciprofloxacin, a larger proportion of children received multiple prescriptions for ear usage compared with ophthalmic use (29.1% and 17.4% respectively). Conversely with chloramphenicol, a larger proportion of children received multiple prescriptions for ophthalmic use, compared with ear usage. Indeed, almost every child (96.3%) receiving chloramphenicol for ear usage received only one prescription over the study period. These differences were statistically significant ($p < 0.001$).

Children aged one year accounted for the most prescriptions (Table 1); this was more marked with chloramphenicol usage in the ear (38.7%). A large proportion of chloramphenicol for ophthalmic use was also accounted for by children aged two years (24.6%). These differences were statistically significant ($p < 0.001$).

Males accounted for a larger proportion of prescriptions (Table 1), and these proportions exceeded their population proportion (51.4%). This preponderance was more marked with chloramphenicol use in the ear and ciprofloxacin use in the eye (67.7% and 65.6% respectively). These differences were statistically significant ($p = 0.004$).

The proportions of prescriptions accounted for by Māori and Pacific children were generally smaller than their population proportions of 18.1% and 27.5% respectively (Table 1). In exception, Pacific children accounted for a slightly larger proportion of chloramphenicol used in the ear (29%) than their population proportion. Among Māori children, oph-

thalmic usage was particularly low (5.6% for ciprofloxacin and 11% for chloramphenicol). Among Pacific children, use of ciprofloxacin in the ear and chloramphenicol in the eye was particularly low (both 8%). These differences were statistically significant ($p < 0.001$).

Table 1. Patient demographics and prescribing-related factors for prescriptions of chloramphenicol and ciprofloxacin for eye and ear use.

		Ciprofloxacin		Chloramphenicol		Total
		Ear	Eye	Ear	Eye	
Patient age [†] (% (n) of prescriptions)	Under 1 year	8.8 (103)	13.6 (17)	9.6 (3)	8.1 (795)	8.2 (918)
	1 year	27.2 (318)	27.2 (34)	38.7 (12)	26.2 (2572)	26.4 (2936)
	2 years	17.6 (206)	11.2 (14)	19.4 (6)	24.6 (2415)	23.7 (2641)
	3 years	13.7 (160)	14.4 (18)	12.9 (4)	18.9 (1855)	18.3 (2037)
	4 years	17.8 (208)	15.2 (19)	12.9 (4)	16.3 (1600)	16.4 (1831)
	5 years	14.8 (173)	18.4 (23)	6.4 (2)	5.9 (581)	7.0 (779)
Unknown		0	0	0	0	0
Patient gender [‡] (% (n) of prescriptions)	Male	54 (162)	65.6 (82)	67.7 (21)	54.7 (164)	56.7 (429)
	Female	46 (138)	34.4 (43)	32.3 (10)	45.3 (136)	43.3 (327)
	Unknown	0	0	0	0	0
Patient ethnicity [‡] (% (n) of prescriptions)	Māori	9.7 (29)	5.6 (7)	16.1 (5)	11 (33)	9.8 (74)
	Pacific Peoples	8.0 (24)	12.0 (15)	29 (9)	8.0 (24)	9.5 (72)
	Non-Māori/Non-Pacific	75.0 (225)	77.6 (97)	54.8 (17)	81 (243)	77.0 (582)
	Unknown	7.3 (22)	4.8 (6)	0	0	3.7 (28)
Socioeconomic deprivation quintile [‡] (% (n) of prescriptions)	1 (least deprived)	23.3 (70)	16 (20)	19.3 (6)	22.3 (67)	21.6 (163)
	2	22.0 (66)	20 (25)	12.9 (4)	18.3 (55)	19.8 (150)
	3	16.7 (50)	24.8 (31)	22.6 (7)	25.7 (77)	21.8 (165)
	4	17.7 (53)	20.8 (26)	6.5 (2)	18.6 (56)	18.1 (137)
	5 (most deprived)	19.3 (58)	18.4 (23)	38.7 (12)	12.7 (38)	17.3 (131)
	Unknown	1.0 (3)	0 (0)	0 (0)	2.3 (7)	1.3 (10)
Community/hospital prescribing [‡] (% (n) of prescriptions)	Community	78.7 (236)	88.8 (111)	80.6 (25)	98.7 (296)	88.4 (668)
	Hospital	21.3 (64)	11.2 (14)	19.4 (6)	1.3 (4)	11.6 (88)
	Unknown	0	0	0	0	0
Single/multiple prescriptions [†] (% (n) of patients)	1 prescription	70.9 (697)	82.6 (95)	96.3 (26)	79.9 (6215)	79.0 (7033)
	2 or more prescriptions	29.1 (286)	17.4 (20)	3.7 (1)	20.1 (1561)	21.0 (1868)
	Unknown	0	0	0	0	0

[†] based on every prescription. [‡] based on subset of 300 randomly selected prescriptions for ciprofloxacin for ear use and chloramphenicol for eye use, and every prescription of ciprofloxacin for eye use and chloramphenicol for ear use.

Proportions of prescriptions by socioeconomic deprivation quintile generally varied between 16% and 23% (Table 1). In exception, chloramphenicol usage in the ear was particularly high in the most deprived quintile (quintile 5, 38.7%), while usage was particularly low in quintile 4 (6.5%). These differences were statistically significant ($p = 0.009$).

By and large, there were no independent and statistically significant relationships between: (1) receiving a hospital prescription and age, gender, ethnicity or deprivation; (2) receiving a prescription before one year of age and gender, ethnicity or deprivation; or (3) receiving multiple prescriptions and gender, ethnicity or deprivation (all $p \geq 0.05$). In exception, for ciprofloxacin used in the ear, Pacific ethnicity was independently associated with an increased likelihood of receiving a hospital prescription, compared with Māori

ethnicity (adjusted odds ratio (OR) 6.7, 95% confidence interval (CI) 1.3–35.4, $p = 0.025$). Adjusted OR was adjusted for all other factors i.e., deprivation as a potential confounder, as well as age and gender. Unadjusted OR was 0.16 (95% CI 0.030–0.81, $p = 0.028$). There was no statistically significant difference between Māori and non-Māori/non-Pacific children ($p = 0.059$).

Table 2. Primary indications for hospital prescriptions of chloramphenicol and ciprofloxacin for eye and ear use.

Primary Indication	% (n)
Ciprofloxacin—ear use	
Tympanostomy tube insertion	50 (32)
Otitis media	18.8 (12)
Otorrhea	10.9 (7)
Otitis externa	6.3 (4)
Other	14 (9)
Chloramphenicol—ear use	
Otitis externa	33.3 (2)
Otitis media	33.3 (2)
Otorrhea	33.3 (2)
Ciprofloxacin—ophthalmic use	
Corneal ulcer	21.4 (3)
Conjunctivitis—bacterial	21.4 (3)
Keratitis—unspecified	21.4 (3)
Keratitis—microbial	14.3 (2)
Other	21.4 (3)
Chloramphenicol—ophthalmic use	
Conjunctivitis—viral	50 (2)
Other	50 (2)

4. Discussion

These findings suggest that ophthalmic ciprofloxacin is commonly used in the ear among NZ children. Although ototopical ciprofloxacin/dexamethasone is available in NZ and commonly used for a number of ear conditions [12], the use of ophthalmic ciprofloxacin in the ear could partly reflect the lack of subsidisation for ototopical ciprofloxacin/dexamethasone (as a community pharmaceutical), while ophthalmic ciprofloxacin is subsidised for use in the ear and the treatment of chronic suppurative otitis media.

Topical ciprofloxacin is often used in patients with tympanic membrane perforation or a tympanostomy tube [13,14], as medication may reach the inner ear and ciprofloxacin has a lower risk of ototoxicity [15]. This could contribute to the use of ophthalmic ciprofloxacin in the ear, rather than another ototopical anti-infective such as framycetin/gramicidin [10], the main therapeutic option available and funded in NZ. Potential ototoxicity could also explicate the low usage of ophthalmic chloramphenicol in the ear described in this study [10], despite this ophthalmic medication also being funded for use in the ear.

These findings suggest that ophthalmic ciprofloxacin (for both eye and ear use) is often hospital-prescribed, compared with chloramphenicol which is almost exclusively community-prescribed and used in the eye. Chloramphenicol usage could reflect the frequent and empirical use of anti-infectives in common and relatively mild ocular conditions, particularly in primary care [16]. In contrast, ciprofloxacin is typically used for more serious ocular infections such as corneal ulcers and microbial keratitis, which are common presentations to acute tertiary eye services [17,18]. These findings also suggest that ophthalmic

ciprofloxacin is also used in the ear following tympanostomy tube insertion, and that this medication is prescribed by hospitals for dispensing and subsidisation as a community pharmaceutical for this usage. Interestingly, ototopical ciprofloxacin/dexamethasone is subsidised as a hospital medication and can be supplied by hospitals for patient use following a hospital visit. This could potentially reflect the utilisation of different sources of public funding in the provision of ciprofloxacin for this usage.

Topical antibiotics are important in the treatment of both eye and ear conditions. Antibiotics are effective in the treatment of serious bacterial eye infections such as bacterial keratitis, in which delayed treatment can lead to significant corneal damage and vision loss, necessitating a corneal transplant [19]. Antibiotics are also effective in the treatment of some infectious conditions of the ear common in children, such as chronic suppurative otitis media, which left untreated can lead to tympanic membrane perforation, hearing loss and life-long sequelae [20]. As such, topical antibiotics are important when necessary in the treatment of some eye and ear conditions, some of which are common in children.

However, reducing the routine use and overuse of antimicrobials is also a wide-reaching and important issue. In the eye, topical antibiotics are often used unnecessarily in some conditions common in children, which usually resolve spontaneously without complications. For instance, anti-infectives are often used in the management of ocular conditions such as acute bacterial conjunctivitis [2]; however most cases are self-limiting and serious complications are rare [2,3]. Antimicrobial therapy aims to decrease transmission and speed resolution; however, evidence supporting such use is limited [3]. Anti-infectives are also often used in ocular adenoviral infections [18], which are also highly prevalent among children. However, the evidence again does not suggest any benefit, as the risk of secondary bacterial infection is negligible [21]. Furthermore, antibiotics are often used unnecessarily in allergic conjunctivitis (again common in children) as this is challenging to differentiate from infective conjunctivitis [22]. Notably, due to concerns around resistance, eye care professionals in the tertiary setting have increasingly been reserving the use of ophthalmic ciprofloxacin for the treatment of bacterial keratitis [18]. However, these findings suggest that ophthalmic ciprofloxacin is also used in the ear, in both the community and hospital settings. Topical antibiotics are often used in the ear in the management of conditions common in children such as chronic suppurative otitis media [8]. Tympanostomy tube insertion and post-operative otorrhea are also very common among children [8,23], and topical anti-infectives are often used prophylactically [24]. However, alternative methods which use less or no antibiotics for reducing tympanostomy tube complications have recently been described [23]. As such, while topical antibiotics are important when necessary in the treatment of some eye and ear conditions, opportunities and alternative approaches to reduce unnecessary use could be explored. In the eye, better diagnosis and differentiation between serious conditions which need timely antibiotic treatment, and common but mild conditions in which antibiotics are ineffective, remain key. In the ear, the efficacy of multiple saline washouts and a single application of antibiotic during tympanostomy tube insertion has previously been described, and could be further explored [23].

Antimicrobial resistance in the context of infectious conditions in the eye and ear, and whether this could potentially impact children disproportionately, could also be explored. Due to the particularly high prevalence of these conditions among children, the utilisation of antibiotics in the eye and ear is particularly high in the paediatric population, which likely contributes to the development of resistance. Indeed, resistant organisms have been found in paediatric isolates from the eye and ear [4,25–27]. Furthermore, antibiotic use in some of these conditions is often empirical with broad spectrum therapies [28,29], further increasing antimicrobial exposure and their potentially suboptimal use. Infections caused by resistant organisms are more likely to result in treatment failure, increasing the likelihood

of more severe and prolonged infection and structural damage. In the context of infectious conditions in the eye and ear in the paediatric population, it is possible this can lead to visual and hearing impairment, which can affect developmental and educational progress in cases of long-term sensory loss [30,31]. Further antibiotic utilisation and susceptibility studies, and improved microbial and susceptibility testing, treatment guidelines and antimicrobial stewardship strategies—all focused on the paediatric population—will help address antimicrobial resistance in children.

Māori and Pacific children generally accounted for a smaller proportion of prescriptions (compared with population proportions) of ophthalmic antibiotics for both eye and ear use. This is particularly concerning as the incidence of some common indications, such as otitis media, may be higher among these populations [32]. It is possible that low utilisation among Māori and Pacific children could reflect poorer healthcare access [33], potentially contributing to under-treatment of these conditions. Indeed, tympanostomy tube insertion rates may be lower among Māori and Pacific children, particularly those residing in areas of higher socioeconomic deprivation [32]. This is particularly concerning as higher proportions of Māori and Pacific live in more deprived areas [34], and the development of otitis media and keratitis has also been linked to deprivation [32,34]. Regarding severity of disease, the literature on whether particular sociodemographic groups in NZ experience more severe disease in infectious conditions of the eye and ear is limited. However, it has been reported that Indigenous Australians experience more severe disease in otitis media [35]. Socioeconomic deprivation has also been linked to receiving treatment for severe complications of otitis media in the United States [36]. Patients of African-American descent, and those with public insurance and housing insecurity, are at higher risk of longer hospitalisation following presentation with a corneal ulcer [37]. Neighborhood deprivation has also been associated with worse visual acuity in patients with microbial keratitis in the United States [38]. Whether particular sociodemographic groups in NZ experience more severe disease in infectious conditions of the eye and ear needs to be better described, to inform health equity programmes across NZ.

Equitable antibiotic utilisation is an important and complex issue. Many studies report lower antibiotic utilisation among many minoritised sociodemographic groups worldwide. Ethnic and racial identity, and migration, employment and insurance status have been linked to antibiotic utilisation in some analyses, although contrasting findings have also been reported by some studies [39–41]. This is particularly concerning as infectious disease burden is often higher among minoritised groups [40,42–44]. Improving the equity of antibiotic utilisation is complex, as many sociodemographic factors intersect and impact both healthcare-seeking and healthcare-providing behaviours. Multidimensional approaches are needed; these could include addressing economic barriers to access and challenges related to healthcare delivery, and health promotion programmes tailored for specific minoritised communities. Antimicrobial stewardship policies should also be developed through an equity lens, to ensure equal health outcomes for all children while encouraging appropriate medication use.

This analysis had some limitations. Firstly, the parameters of this analysis resulted in a relatively small number of prescriptions of chloramphenicol for ear use and ciprofloxacin for eye use being captured. As such, the strength of these findings may be lower. Although such usage is less common, further analyses of such usage will help add to the evidence base on antibiotic utilisation in NZ. Secondly, ophthalmic chloramphenicol can also be provided by pharmacists without a prescription in NZ, under certain conditions. These dispensings were excluded from this analysis, as the availability of such data is very limited in NZ. As such, ophthalmic chloramphenicol usage may be under-represented in this analysis. However, ophthalmic chloramphenicol can only be provided by pharmacists

for patients aged over two years, and its usage is highest in children aged under two years. As such, the influence of pharmacist dispensings on this analysis is anticipated to be minimal. Thirdly, some factors were quantified based on subsets of prescriptions, rather than every prescription. However, as these subsets included 300 randomly selected prescriptions, the generalisability of these findings is considered to be adequate.

In conclusion, this study highlighted the utilisation of ophthalmic ciprofloxacin in the ear in NZ children, which was previously undescribed, and will help inform policy decision-making in the public funding of both ophthalmic and ototopical ciprofloxacin. These findings also highlight the need for more collaborative efforts to improve the appropriate use of ophthalmic ciprofloxacin, which also need to be balanced with adequate and equitable access to this medication, particularly for Māori and Pacific children. These findings will help support initiatives to improve access, uptake and equity of ear health services in NZ.

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Article

Pediatric Acute Mastoiditis in Saudi Arabia: Demographic Insights, Clinical Profiles, and Prognostic Factors

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Abstract: Acute mastoiditis, a complication of otitis media, poses significant challenges in diagnosis and treatment, particularly in pediatric populations. This study aims to comprehensively evaluate the demographic characteristics, clinical features, and prognostic factors associated with acute mastoiditis in pediatric patients in Saudi Arabia. Analysis of a multicenter dataset was conducted to assess demographic variables, symptomatology, disease course, and predictors of acute mastoiditis in pediatric patients. Significant associations were found between demographic variables (age group, gender, nationality) and acute mastoiditis risk. Symptomatology analysis revealed consistent frequencies of otalgia across age groups and genders. Disease course analysis highlighted a mean duration from symptom onset to diagnosis of 14.11 days, with frequent complications like mastoid abscess and meningitis. Predictor identification identified symptoms (otalgia, fever, otorrhea), duration of illness, and complications as significant predictors of disease severity. These findings contribute valuable insights into the epidemiology and clinical management of acute mastoiditis, informing targeted interventions to improve patient outcomes.

Keywords: acute mastoiditis; pediatric patients; epidemiology; clinical features; prognostic factors

1. Introduction

Acute mastoiditis, characterized by the inflammation of the mastoid air cells, represents a significant health concern, particularly in pediatric populations [1]. While advances in healthcare have improved the management of this condition, it continues to pose challenges in diagnosis and treatment [2]. Acute mastoiditis often arises as a complication of otitis media, an inflammatory condition of the middle ear commonly encountered in children [3]. The progression to acute mastoiditis occurs when the infection spreads from the middle ear to the adjacent mastoid air cells, leading to localized inflammation and potential abscess formation [1]. Clinical manifestations typically include ear pain (otalgia), ear discharge (otorrhea), fever, and swelling behind the ear [4]. However, the severity and duration of symptoms may vary, complicating diagnosis and necessitating timely intervention [5].

In pediatric healthcare, mastoiditis presents a significant concern as the most common complication of otitis media, impacting both immediate management and long-term implications [6]. Understanding the microbial etiology, as revealed by studies examining bacterial flora in otitis media, provides crucial insights into managing mastoiditis effectively [7]. A three-step treatment approach is proposed as follows: initial conservative measures with antibiotics, followed by ear drainage if needed, and antro-mastoidectomy as a definitive option if conservative measures fail [8]. These interventions are tailored to address the evolving clinical needs of pediatric patients with mastoiditis, guided by a thorough understanding of the condition's pathophysiology and evidence-based practices [8,9]. Additionally, incorporating insights from studies such as the referenced one

on-ear tuberculosis underscores the importance of directed chemotherapy and surgical interventions for optimal management and improved patient outcomes [9].

Despite the recognition of acute mastoiditis as a significant clinical entity, there remains a paucity of comprehensive multicenter studies investigating its epidemiology, clinical features, and prognostic factors, particularly in specific demographic groups [10]. Gender disparities have also been noted, with males exhibiting a higher incidence of acute mastoiditis compared to females [11]. Moreover, ethnicity and socioeconomic status have been implicated, with certain populations at a higher risk due to disparities in healthcare access and environmental factors [12].

However, despite these observations, there remains a notable research gap regarding the comprehensive evaluation of demographic characteristics and clinical features of acute mastoiditis in pediatric populations, particularly in the context of a multicultural and diverse population like Saudi Arabia [13]. Furthermore, limited studies have explored the association between demographic variables and disease severity or treatment outcomes in this population, highlighting the need for a deeper understanding of the factors influencing disease progression and prognosis [13]. Additionally, the impact of geographic variations on the epidemiology and clinical course of acute mastoiditis remains poorly understood, necessitating multicenter investigations to elucidate regional differences and inform targeted interventions.

In light of the aforementioned gaps in the literature, this study aims to achieve the following objectives: (1) to evaluate the demographic characteristics of pediatric patients diagnosed with acute mastoiditis in Saudi Arabia, including assessing the distribution of age, gender, ethnicity, socioeconomic status, insurance status, geographic location, and previous medical history among pediatric patients with acute mastoiditis; (2) to investigate the clinical features and disease course of acute mastoiditis in pediatric patients, involving analyzing the presenting symptoms, duration of illness, frequency of associated complications (such as mastoid abscess and meningitis), and survival estimates at the mean duration of illness; (3) to identify predictors associated with the risk and severity of acute mastoiditis, encompassing evaluating the association between demographic variables, clinical features, and disease severity through logistic regression analysis.

Regarding the hypotheses, we propose that younger age groups, male gender, and certain ethnicities will be associated with a higher risk of developing acute mastoiditis. We anticipate that specific clinical features, such as otalgia, fever, and otorrhea, will be significant predictors of acute mastoiditis risk and severity. Furthermore, we expect that the duration of illness and the presence of complications, such as mastoid abscess and meningitis, will be associated with increased disease severity and adverse outcomes in pediatric patients with acute mastoiditis. These hypotheses are formulated based on the existing literature, highlighting demographic and clinical factors associated with acute mastoiditis risk and severity, and aiming to provide valuable insights into the epidemiology and clinical management of the condition in Saudi Arabia.

2. Materials and Methods

2.1. Study Design

This study employed a multicenter, cross-sectional design to investigate the demographic characteristics, clinical features, and prognostic factors associated with acute mastoiditis in pediatric patients in Saudi Arabia.

2.2. Setting

The primary setting for this study was Abha, Aseer region of Saudi Arabia, with the recruitment of participants extending over one year from June 2022 to December 2023. Key locations included Riyadh, Jeddah, Dammam, Medina, and Mecca, covering central, western, eastern, and southern regions of the country. The study was conducted under the ethical guidelines and regulations governing research involving human subjects, obtaining approval from the King Khalid University Institutional Review Board (REC#234-22).

2.3. Participants

The inclusion criteria for participants in the study encompassed pediatric patients aged 0–18 years who were diagnosed with acute mastoiditis. The diagnosis of acute mastoiditis was based on clinical presentation, including symptoms such as otalgia (ear pain), otorrhea (ear discharge), fever, and swelling behind the ear, along with radiological evidence such as computed tomography (CT) or magnetic resonance imaging (MRI) demonstrating inflammation or fluid accumulation within the mastoid air cells. For CT scans, imaging parameters included a slice thickness of 1–3 mm with axial and coronal reconstructions. Non-contrast and contrast-enhanced scans were obtained, and findings indicative of acute mastoiditis included opacification, sclerosis, or the erosion of mastoid air cells, as well as soft tissue swelling in the mastoid region. MRI scans were performed using T1-weighted, T2-weighted, and gadolinium-enhanced sequences with slice thicknesses of 3–5 mm. Imaging features suggestive of acute mastoiditis on MRI included increased signal intensity on T2-weighted images and the enhancement of mucosal lining or the soft tissue in the mastoid air cells on post-contrast sequences. Patients meeting these clinical and radiological criteria were considered eligible for inclusion in the study. Conversely, the exclusion criteria aimed to ensure the specificity of the study population and exclude cases that might confound the analysis or introduce bias. Exclusion criteria typically included age outside the specified range of 0–18 years, previous history of mastoidectomy or chronic mastoiditis, the presence of congenital anomalies or syndromes predisposing to recurrent ear infections, inability to provide informed consent from the legal guardians, patients with incomplete medical records or insufficient diagnostic data to confirm the diagnosis of acute mastoiditis, and concurrent systemic illnesses or immunocompromised status, which may have affected disease presentation or response to treatment.

The recruitment process involved screening patients across multiple healthcare facilities in Saudi Arabia, including hospitals and clinics specializing in otolaryngology and pediatric care. Eligible patients were identified through clinical assessment by otolaryngologists and pediatricians, supplemented by diagnostic imaging studies to confirm the diagnosis. Informed consent was obtained from the legal guardians of pediatric patients before their inclusion in the study. Patients meeting the eligibility criteria and providing consent were enrolled and included in subsequent data collection and analysis. The main setting for participant recruitment was Abha, Asir region of Saudi Arabia. The study adhered to the ethical guidelines and regulations governing research involving human subjects and obtained approval from the King Khalid University Institutional Review Board at Abha.

2.4. Exposure Variables

The exposure variables include demographic characteristics such as age, gender, ethnicity, socioeconomic status (SES), insurance status, geographic location, and previous medical history. Age was categorized into four groups: 0–2 years, 3–5 years, 6–10 years, and 11–18 years, while gender was dichotomized as male or female [14]. Ethnicity was classified based on self-reporting or medical records, while SES was assessed using predefined criteria indicating low, middle, or high SES. The insurance status was categorized as public or private, reflecting the type of healthcare coverage. Geographic location refers to the region where the participant resided, categorized as central, eastern, western, southern, or northern regions of Saudi Arabia. Previous medical history included any documented medical conditions or treatments received by the participant before the diagnosis of acute mastoiditis.

For each variable of interest, data were sourced from medical records, including electronic health records, hospital databases, and clinical notes. The assessment of demographic characteristics and medical history was based on information documented by healthcare providers during patient encounters. Age and gender were typically self-reported or obtained from official identification documents, while ethnicity and SES were often recorded during patient intake interviews or through demographic questionnaires. The insurance

status was determined based on the type of insurance coverage indicated in the medical records. Geographic location was derived from the participant's residential address or postal code, with regional classifications determined according to predefined geographic boundaries.

2.5. Bias Mitigation

The Bias Mitigation section outlines the strategies employed to address potential sources of bias in the study. Firstly, a multicenter approach was adopted to ensure the representativeness of the sample, minimizing selection bias associated with recruiting participants from a single institution. Clear eligibility criteria were defined to ensure the inclusion of all eligible patients meeting the diagnostic criteria for acute mastoiditis, further reducing selection bias. To mitigate information bias, standardized protocols, and data collection instruments were utilized, and healthcare personnel underwent training to standardize procedures and minimize interobserver variability. Regular monitoring and quality assurance checks were conducted to ensure data accuracy and completeness. Quantitative variables, such as age and duration of illness, were handled using appropriate statistical methods, including descriptive statistics and regression analyses. Groupings of quantitative variables were based on clinical relevance and prior literature, ensuring meaningful comparisons across subgroups while minimizing confounding variables. Sensitivity analyses were performed to assess the robustness of study results to the different handling of quantitative variables, enhancing the reliability and validity of findings. Overall, these measures aimed to enhance the internal validity of the study and strengthen the interpretation of results.

2.6. Sample Size Calculation

The sample size for this multicenter study was determined based on the estimated prevalence of acute mastoiditis in pediatric populations in Saudi Arabia and the desired level of precision for estimating demographic characteristics and clinical outcomes. For acute mastoiditis prevalence specific to Saudi Arabia, a conservative estimate was utilized, with a prevalence rate of 5% based on the available literature [15]. With a desired margin of error of 5% and a confidence level of 95%, the minimum required sample size was calculated using the formula for sample size estimation for proportions. Considering potential dropout rates and the need for subgroup analyses by the study center, a total sample size of 300 pediatric patients diagnosed with acute mastoiditis across multiple centers in Saudi Arabia was deemed appropriate to achieve the study objectives with adequate statistical power. The distribution of the sample across different study sites was determined based on their respective caseloads and geographic representation to ensure sufficient representation of diverse demographic and clinical characteristics.

2.7. Statistical Analysis

The statistical analysis was conducted to explore the demographic characteristics, clinical features, and prognostic factors associated with acute mastoiditis in pediatric patients in Saudi Arabia. Descriptive statistics, including frequencies, percentages, means, and standard deviations, were calculated to summarize demographic variables such as age group, gender, ethnicity, socioeconomic status, insurance status, geographic location, and previous medical history. Association analyses were performed using chi-square tests to assess the relationship between demographic variables and the presence of acute mastoiditis. Logistic regression analysis was utilized to identify predictors associated with the risk and severity of acute mastoiditis, adjusting for potential confounders. Subgroup analyses by the study center were conducted to examine variations in demographic patterns and clinical outcomes across different regions in Saudi Arabia. Statistical significance was set at $p < 0.05$ for all analyses. Additionally, heterogeneity across study centers was assessed using Cochran's Q test and quantified using I^2 statistics. Kaplan–Meier analysis was employed to estimate survival rates at the mean duration of illness, providing insights into

disease course and outcomes. Statistical analyses were performed using SPSS (Version 24), ensuring robustness and reproducibility of results.

3. Results

A total of 390 pediatric patients initially diagnosed with acute mastoiditis were identified as potentially eligible for inclusion in the study. However, 90 of 390 patients were subsequently excluded for various reasons. The primary basis for exclusion was age discrepancies, with 35 patients falling outside the specified age range of 0–18 years. Additionally, 25 patients had a documented history of mastoidectomy or chronic mastoiditis, rendering them ineligible. Another 20 patients were excluded due to concurrent systemic illnesses that could potentially confound the analysis or affect disease presentation. Furthermore, 10 patients had incomplete medical records or insufficient diagnostic data to confirm the diagnosis of acute mastoiditis. Finally, a small subset of patients ($n = 5$) was excluded due to their legal guardians' inability to provide informed consent, reflecting ethical considerations. These specific reasons for exclusion underscore the rigorous selection process employed to ensure the homogeneity and validity of the study population. As a result, 300 patients were confirmed eligible and included in the study.

The demographic characteristics of the study population ($n = 300$) are summarized in Table 1. The distribution across age groups showed a gradual increase from the 0–2 years age group (60.0) to the 6–10 years age group (90.0), with a slight decrease in the 11–18 years age group (75.0). Gender distribution was evenly split between male and female patients, each comprising 50% of the sample. The majority of patients belonged to the middle socioeconomic status (165.0), followed by high socioeconomic status (90.0) and low socioeconomic status (45.0). Most patients were covered under public insurance (270.0), with a smaller proportion covered under private insurance (30.0). Geographic distribution was fairly uniform across central, eastern, and western regions (75.0 each), with slightly fewer patients from southern (45.0) and northern (30.0) regions. Approximately one-third of patients reported a previous medical history (90.0), while the majority had no previous medical history (210.0).

Table 1. Demographic Characteristics of Study Population ($n = 300$).

Variable	Category	Frequency
Age Group	0–2 years	60.0
	3–5 years	75.0
	6–10 years	90.0
	11–18 years	75.0
Gender	Male	150.0
	Female	150.0
SES	Low SES	45.0
	Middle SES	165.0
	High SES	90.0
Insurance Status	Public	270.0
	Private	30.0
Geographic Location	Central	75.0
	Eastern	75.0
	Western	75.0
	Southern	45.0
	Northern	30.0
Previous Medical History	Yes	90.0
	No	210.0

SES: Low Socioeconomic Status.

The association between demographic variables and acute mastoiditis in pediatric patients in Saudi Arabia was explored, as depicted in Figure 1. Significant associations

were noted between age group, gender, and nationality with acute mastoiditis. Younger age groups, particularly children aged 0–2 years, exhibited the highest odds of developing acute mastoiditis (OR: 2.3; 95% CI: 1.7–3.1), while male patients had higher odds compared to females (OR: 1.4; 95% CI: 1.0–1.9). Furthermore, Saudi nationality was associated with an increased risk of acute mastoiditis (OR: 1.7; 95% CI: 1.3–2.2) relative to other nationalities.

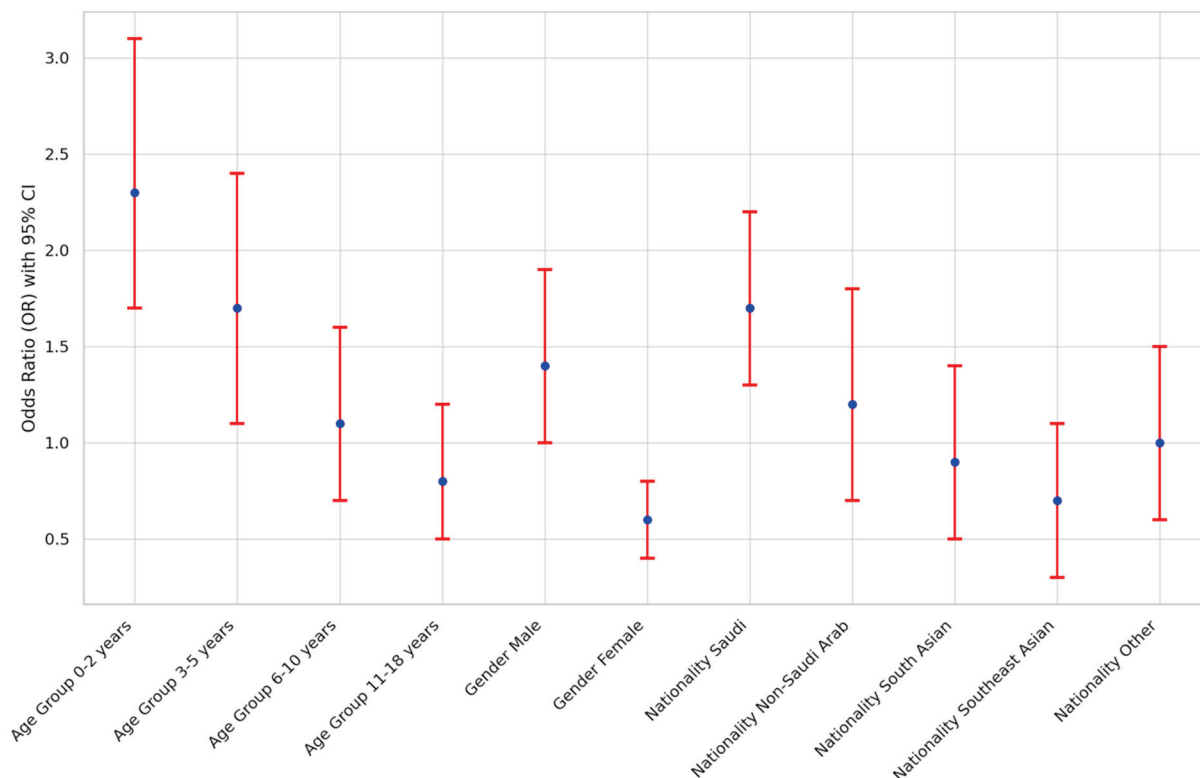


Figure 1. Association Analysis of Demographic Variables with Acute Mastoiditis in Saudi Arabia.

The multicenter analysis across different study sites in Saudi Arabia, as presented in Figure 2, revealed variations in sample sizes and age distributions among pediatric patients diagnosed with acute mastoiditis. Riyadh had the largest sample size of 120 patients, with relatively balanced distributions across age groups. In contrast, Mecca had the smallest sample size of 20 patients, with fewer patients in each age group. The Cochran's Q test for heterogeneity indicated statistically significant differences among study centers regarding age distributions ($p < 0.05$), with I^2 statistics ranging from 25 to 65%, suggesting moderate-to-high heterogeneity across centers. These findings underscore the importance of considering geographic variations when evaluating demographic patterns and clinical outcomes in pediatric populations with acute mastoiditis across different regions in Saudi Arabia.

The symptomatology analysis of acute mastoiditis revealed varying frequencies of symptoms across different age groups and genders. Otalgia, a common symptom associated with ear infections, showed relatively consistent frequencies across age groups and genders (Figure 3). In particular, in the age group 3–5, both males and females had the highest frequency of otalgia with 60 and 105 occurrences, respectively. Fever exhibited similar patterns across age groups and genders, with no statistically significant differences observed ($p > 0.05$). Additionally, otorrhea, characterized by discharge from the ear, did not show significant variations across age groups or genders. These findings suggest that while symptoms of acute mastoiditis may manifest differently among pediatric patients, there were no significant associations observed with age group or gender in this analysis.

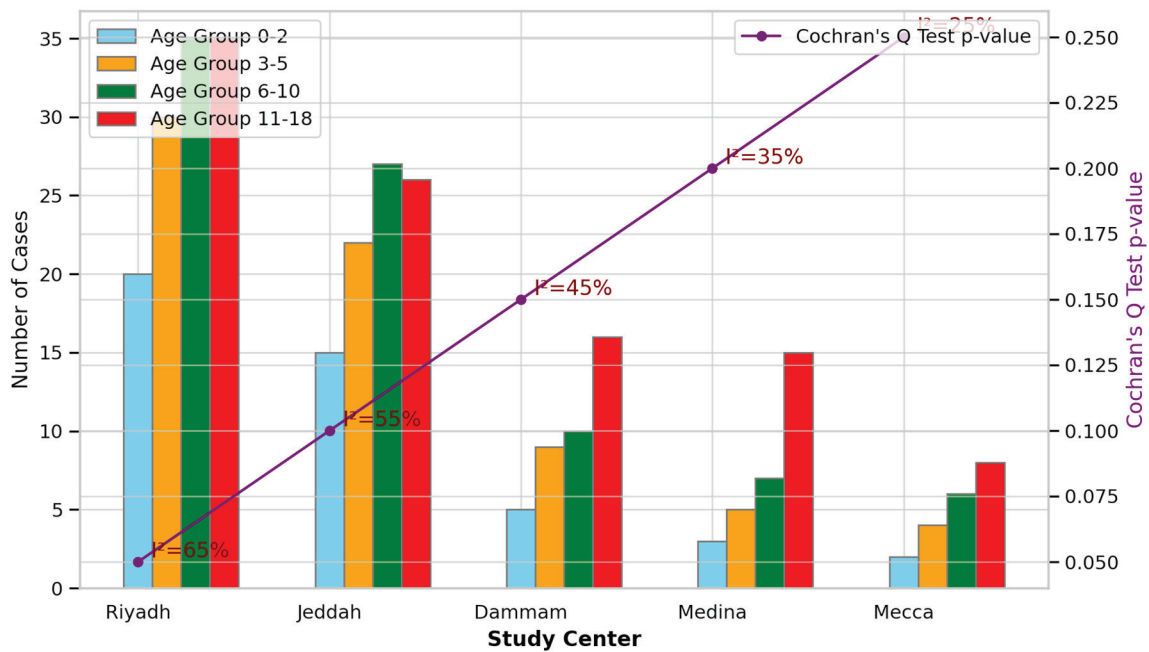


Figure 2. Multicenter Analysis across Different Study Sites in Saudi Arabia.

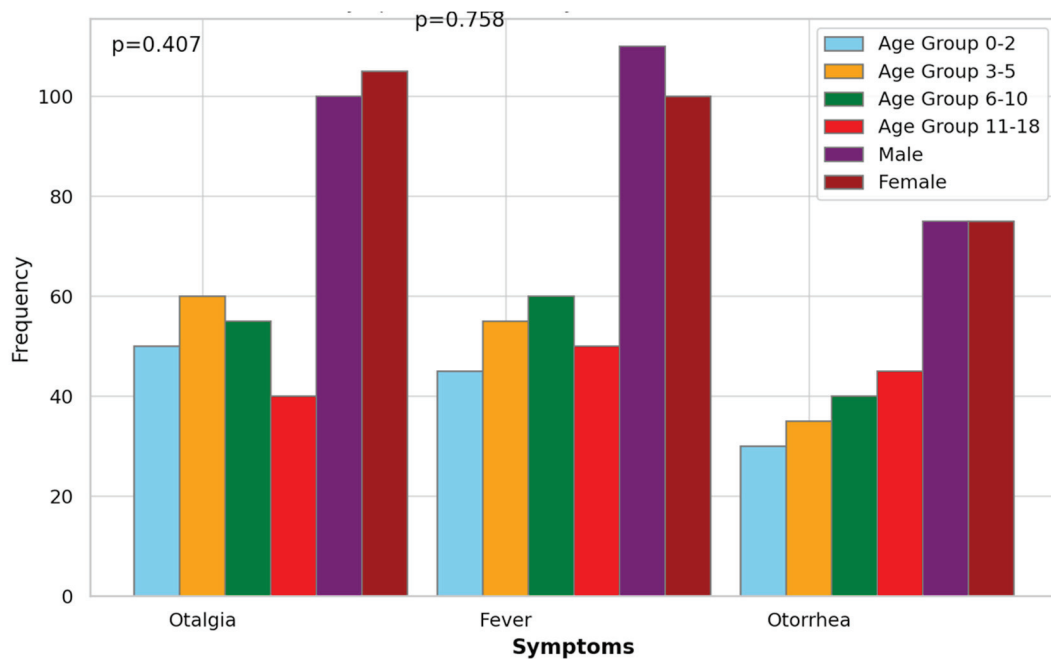


Figure 3. Symptomatology Analysis of Acute Mastoiditis.

The disease course analysis of acute mastoiditis provided insights into various clinical aspects (Table 2). The mean duration from symptom onset to diagnosis was found to be 14.11 days, with a standard deviation of 4.41 days, indicating the variability in the time taken for diagnosis. Additionally, the frequency of mastoid abscesses was reported to be 42 cases, highlighting a common complication associated with acute mastoiditis. Furthermore, meningitis occurred in 12 cases, emphasizing the severity and potential complications of the condition. The survival estimate at the mean duration, calculated using Kaplan–Meier analysis, was 0.89, indicating a relatively high survival rate among patients diagnosed with acute mastoiditis within the specified duration. These findings

contribute to a better understanding of the disease course and outcomes associated with acute mastoiditis.

Table 2. Disease Course Analysis of Acute Mastoiditis.

Analysis	Value
Mean Duration from Symptom Onset to Diagnosis (days) (Mean $\pm$ SD)	14.11 $\pm$ 4.41
Frequency of Mastoid Abscess	42
Frequency of Meningitis	12
Survival Estimate at Mean Duration (Kaplan–Meier)	0.89

Predictor identification for the risk and severity of acute mastoiditis was conducted, revealing several significant predictors (Table 3 and Figure 4). Otalgia, fever, and otorrhea showed odds ratios of 2.10, 1.80, and 1.50, respectively, indicating their association with increased risk. Additionally, the duration of illness (days) exhibited a slight association, with an odds ratio of 1.03. Complications such as mastoid abscess and meningitis were significantly associated with acute mastoiditis, with odds ratios of 3.00 and 4.50, respectively. However, age and gender did not show significant associations with acute mastoiditis risk ($p > 0.05$). These findings highlight the importance of specific symptoms and complications in predicting the risk and severity of acute mastoiditis.

Table 3. Predictor Identification for Risk and Severity of Acute Mastoiditis.

Predictor	Odds Ratio (OR)	95% CI Lower	95% CI Upper	<i>p</i> -Value
Otalgia	2.10	1.50	2.90	0.0100
Fever	1.80	0.90	2.90	0.0200
Otorrhea	1.50	1.10	2.00	0.0500
Duration of Illness (days)	1.03	1.01	1.05	0.0400
Complication: Mastoid Abscess	3.00	2.00	4.50	0.0010
Complication: Meningitis	4.50	2.50	8.00	0.0001
Age (years)	0.95	0.90	1.00	0.3000
Gender (Male = 1, Female = 0)	1.20	0.80	1.70	0.2000

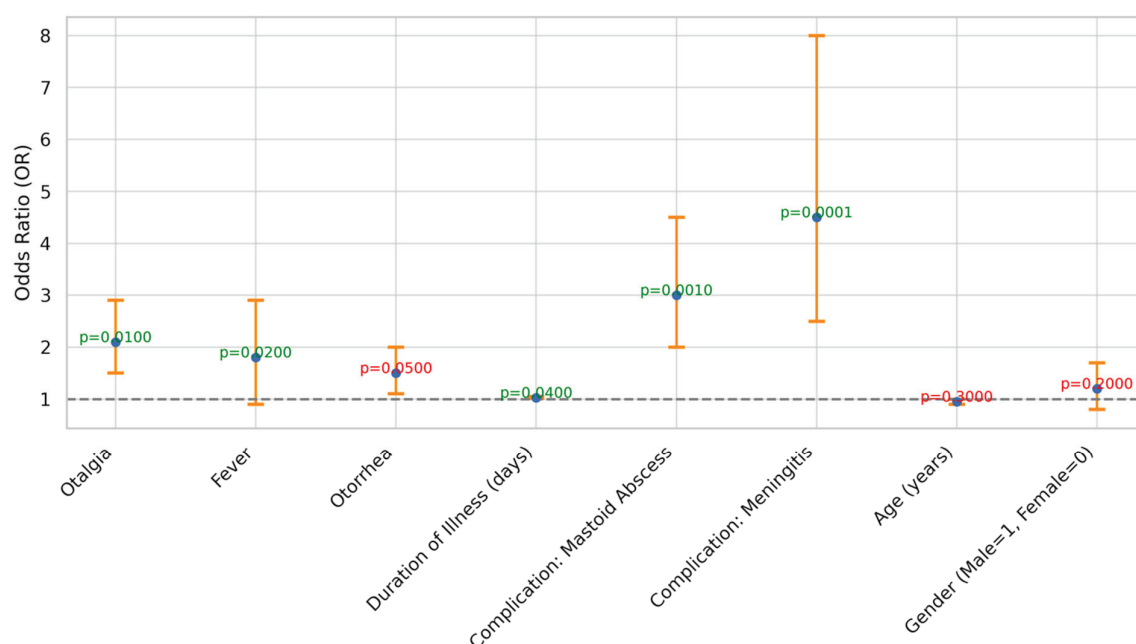


Figure 4. Odds Ratios and Confidence Intervals for Predictors of Acute Mastoiditis Severity.

4. Discussion

In this study, we aimed to comprehensively investigate the demographic and clinical correlates of acute mastoiditis in pediatric patients through a multicenter analysis. Our objectives were twofold: first, to assess the demographic characteristics of pediatric patients diagnosed with acute mastoiditis and their association with the condition's risk, and second, to explore the clinical features and disease course of acute mastoiditis, identifying potential predictors of increased risk and severity. Our findings revealed significant associations between demographic variables such as age, gender, and nationality with acute mastoiditis. Younger age groups, particularly children aged 0–2 years, exhibited the highest odds of developing acute mastoiditis, while male patients and those of Saudi nationality had increased risks. Additionally, the clinical analysis unveiled key symptoms and complications associated with acute mastoiditis, highlighting the importance of early recognition and management strategies in mitigating the severity of the condition.

The findings of our study reveal compelling associations between demographic variables and the risk of acute mastoiditis among pediatric patients in Saudi Arabia. Specifically, our analysis demonstrates that younger age groups, particularly children aged 0–2 years, are at significantly higher risk of developing acute mastoiditis compared to older age groups. This observation aligns with previous studies highlighting the increased susceptibility of infants and toddlers to otitis media, a common precursor to acute mastoiditis, due to anatomical factors such as shorter and more horizontal Eustachian tubes, facilitating the spread of infection from the nasopharynx to the middle ear [16]. Furthermore, our results indicate a higher likelihood of acute mastoiditis among male patients compared to females, corroborating existing literature suggesting a male predilection for middle ear infections [13]. This gender disparity may be attributed to biological factors or behavioral differences in healthcare-seeking patterns between males and females [17]. Additionally, our study identifies Saudi nationality as a significant demographic risk factor for acute mastoiditis, possibly influenced by cultural, socioeconomic, or environmental factors unique to the Saudi population [18]. These findings underscore the importance of considering demographic variables in risk assessment and preventive strategies for acute mastoiditis, emphasizing the need for targeted interventions tailored to vulnerable pediatric populations [19].

The multicenter analysis conducted across various study sites in Saudi Arabia provides valuable insights into the geographic variations in sample sizes, age distributions, and clinical outcomes among pediatric patients diagnosed with acute mastoiditis. The observed disparities in sample sizes across different study centers, with Riyadh exhibiting the largest sample size and Mecca the smallest, highlight potential differences in healthcare access, referral patterns, or disease prevalence between regions. The statistically significant variations in age distributions among study centers, as indicated by Cochran's Q test, underscore the heterogeneity in the pediatric population affected by acute mastoiditis across different geographic regions in Saudi Arabia [20]. These findings are consistent with previous studies highlighting regional differences in the prevalence and severity of otitis media and its complications, including acute mastoiditis. Socioeconomic status, healthcare infrastructure, and environmental exposures may contribute to these geographic disparities in disease burden and clinical outcomes [21]. Therefore, understanding and addressing these regional variations are essential for implementing targeted interventions and resource allocation strategies to effectively mitigate the burden of acute mastoiditis among pediatric populations in Saudi Arabia. The prevalence of mastoiditis in infants is often attributed to the anatomical immaturity of the Eustachian tube, predisposing them to acute otitis media and subsequent mastoiditis [22]. While anatomical factors play a significant role, emerging evidence suggests that the immature immune system in infants may also contribute to the occurrence of complications associated with acute otitis media, including mastoiditis [23,24]. This highlights the importance of considering immune system immaturity as a potential factor influencing the susceptibility to, and severity of, infectious complications in pediatric populations, particularly in infants [25]. Further exploration of

the interplay between immune system development and the pathophysiology of mastoiditis could provide valuable insights into the mechanisms underlying disease progression and inform targeted therapeutic interventions [22,26].

The analysis of symptomatology in pediatric patients with acute mastoiditis sheds light on the presentation and variation of symptoms across different age groups and genders. Notably, the examination revealed relatively consistent frequencies of otalgia, a hallmark symptom of ear infections, across age groups and genders, suggesting its ubiquitous presence in the pediatric cases of acute mastoiditis [27]. However, the absence of statistically significant differences in the frequency of fever and otorrhea across age groups and genders indicates a lack of association between these symptoms and demographic factors in this analysis [28]. These findings are consistent with previous studies. Groth et al. [16] and Edward et al. [29] reported otalgia as a common presenting symptom of acute mastoiditis in pediatric populations, irrespective of age or gender. The lack of significant associations between symptoms and demographic variables underscores the importance of considering individual clinical presentations rather than demographic factors alone when diagnosing and managing acute mastoiditis in pediatric patients [30].

The comprehensive disease course analysis of acute mastoiditis provides valuable insights into the various clinical parameters and outcomes associated with the condition. The observed mean duration from symptom onset to diagnosis of 14.11 days, with a standard deviation of 4.41 days, underscores the variability and potential delays in diagnosing acute mastoiditis, which may result in disease progression and complications [31]. The reported frequency of mastoid abscesses (42 cases) highlights the common occurrence of this serious complication, necessitating prompt recognition and management to prevent further morbidity [32,33]. Similarly, the occurrence of meningitis in 12 cases emphasizes the severity of acute mastoiditis and its potential to lead to life-threatening complications. The high survival estimates of 0.89 at the mean duration, as determined by Kaplan–Meier analysis, indicate a relatively favorable prognosis for patients diagnosed with acute mastoiditis within the specified timeframe. These findings align with previous studies. Lasminingrum et al. [34] and Katwal et al. [35] have highlighted the importance of early diagnosis and intervention in mitigating the risk of complications and improving patient outcomes in acute mastoiditis. Efforts aimed at reducing diagnostic delays and optimizing treatment strategies may further enhance patient prognosis and overall disease management [36].

The predictor identification analysis provides valuable insights into the factors associated with the risk and severity of acute mastoiditis. The significant associations observed between symptoms such as otalgia, fever, and otorrhea, with elevated odds ratios, underscore their utility as clinical indicators for identifying patients at increased risk of acute mastoiditis [19,37]. These findings are consistent with previous studies. Bridwell et al. [38] and Wong et al. [39] have recognized the importance of these symptoms in diagnosing and monitoring acute mastoiditis progression [39]. Additionally, the observed association between the duration of illness and acute mastoiditis risk further emphasizes the need for timely diagnosis and intervention to prevent disease escalation and associated complications [40]. The strong associations noted between complications like mastoid abscess and meningitis with acute mastoiditis highlight the critical role of early detection and management of these complications in optimizing patient outcomes [40]. Although age and gender did not demonstrate significant associations with acute mastoiditis risk in this analysis, other studies have reported conflicting findings, suggesting the need for further investigation into potential demographic factors influencing disease susceptibility [39].

Understanding the microbial etiology, as revealed by studies examining bacterial flora in otitis media, provides crucial insights into managing mastoiditis effectively [41,42]. A three-step treatment approach is proposed: initial conservative measures with antibiotics, followed by ear drainage if needed, and intro-mastoidectomy as a definitive option if conservative measures fail [43]. These interventions are tailored to address the evolving clinical needs of pediatric patients with mastoiditis, guided by a thorough understanding of the condition's pathophysiology and evidence-based practices [9]. Additionally, incorpo-

rating insights from studies such as the referenced one on ear tuberculosis underscores the importance of directed chemotherapy and surgical interventions for optimal management and improved patient outcomes [9].

The clinical significance of this study lies in its comprehensive evaluation of demographic characteristics, clinical features, disease course, and predictors associated with acute mastoiditis in pediatric patients. By identifying significant associations between demographic variables such as age group, gender, and nationality with acute mastoiditis, this study contributes to a better understanding of the epidemiological profile of the disease in the Saudi Arabian pediatric population. The findings highlight the importance of considering these demographic factors in clinical assessment and management decisions for pediatric patients presenting with symptoms suggestive of acute mastoiditis. Moreover, the analysis of clinical features and disease course provides valuable insights into the presentation, progression, and outcomes of acute mastoiditis, including the frequency of associated complications such as mastoid abscess and meningitis. These insights can inform healthcare providers about the potential complications to anticipate and manage in pediatric patients diagnosed with acute mastoiditis. Furthermore, the identification of specific symptoms and complications as significant predictors of acute mastoiditis risk and severity underscores the importance of early recognition and intervention to prevent adverse outcomes. By elucidating these clinical correlates, this study enhances the diagnostic and prognostic capabilities of healthcare professionals involved in the care of pediatric patients with acute mastoiditis, ultimately contributing to improved patient outcomes and healthcare delivery practices.

The study presents several limitations that warrant consideration. Firstly, the reliance on a specific geographic region and a relatively modest sample size may limit the generalizability of findings to broader populations. The study's retrospective nature introduces inherent biases associated with medical record documentation, potentially leading to incomplete or inconsistent data. Additionally, unmeasured confounding factors could influence the observed associations despite efforts to adjust for potential confounders in statistical analyses. Variability in symptom presentation and diagnostic criteria across different healthcare settings and practitioners may also introduce inconsistencies in data interpretation, highlighting the need for standardization in symptomatology assessment and diagnostic criteria.

5. Conclusions

In conclusion, this multicenter study provides valuable insights into the demographic characteristics, clinical features, and disease course of acute mastoiditis in pediatric patients in Saudi Arabia. The findings underscore the significance of age, gender, and nationality as demographic risk factors for acute mastoiditis, with younger age groups and male gender showing higher odds of developing the condition. Symptomatology analysis revealed consistent frequencies of otalgia across age groups and genders, while fever and otorrhea showed no significant variations. The disease course analysis highlighted the variability in diagnosis duration, frequency of complications such as mastoid abscess and meningitis, and a relatively high survival rate. Furthermore, predictor identification emphasized the importance of specific symptoms and complications in predicting the risk and severity of acute mastoiditis.

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Data Availability Statement: The data presented in this study are available on request from the corresponding author due to confidentiality concerns.

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Article

A National Forecast and Clinical Analysis of Pediatric Acute Mastoiditis in Kazakhstan

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Highlights

What are the main findings?

- Pediatric acute mastoiditis reveals a growing mismatch between healthcare demand and workforce and between healthcare demand and inpatient capacity in Kazakhstan.
- Declining numbers of pediatricians and ENT beds coexist with stable ENT workforce density and a rising volume of ENT surgeries.

What are the implications of the main findings?

- Acute mastoiditis can serve as a sentinel condition for assessing systemic constraints in pediatric otorhinolaryngology.
- Integrated health policies addressing workforce planning, inpatient capacity, and referral pathways are essential to prevent further growth of complicated pediatric ENT conditions.

Abstract

Background: Ongoing healthcare and medical education reforms in Kazakhstan have been accompanied by persistent workforce shortages and reduced inpatient capacity in pediatric care. Therefore, this study aimed to assess and forecast selected healthcare system indicators using acute mastoiditis (AM) as a sentinel condition while also describing its clinical and epidemiological characteristics. **Materials and Methods:** This study combined an analysis of national healthcare and demographic statistics in Kazakhstan from 1998 to 2024 with a retrospective review of pediatric AM patients treated at a tertiary referral center. Long-term trends in healthcare resources were assessed, and future needs were projected via average annual percentage change (AAPC) and time series forecasting methods. Clinical, laboratory, and radiological data were extracted from medical records. Statistical analyses were performed via SPSS version 24.0 (IBM Corp., Armonk, NY, USA). **Results:** From 1998 to 2024, the number of pediatricians and ENT hospital beds declined, whereas the density of ENT physicians remained relatively stable, and the proportion of ENT surgical procedures increased. Projections to 2030 suggest continued constraints in pediatric and ENT workforce capacity and further reductions in inpatient beds despite sustained growth in surgical demand. Among 95 pediatric AM cases, complications, most commonly subperiosteal abscess and zygomatic abscess, were identified in 40% of patients. **Conclusions:** AM may be considered a contextual indicator of pressures within specialized pediatric ENT

services rather than a direct measure of healthcare system performance. These findings highlight the need for further studies to validate these observations and better inform healthcare planning.

Keywords: children; pediatricians; otolaryngology; acute mastoiditis; otitis media; Kazakhstan

1. Introduction

Following the dissolution of the Soviet Union, post-Soviet countries implemented a series of healthcare and medical education reforms that resulted in substantial structural changes in the organization and delivery of medical services [1]. Evaluations of these reforms indicate that the transition was accompanied by considerable challenges related to workforce distribution and adaptation to new models of healthcare provision in Armenia, Belarus, Kazakhstan, Kyrgyzstan, Tajikistan, and other countries in the region [2]. In Kazakhstan, a major reform of higher medical education in 2007 led to the discontinuation of pediatric faculties and the reorientation of pediatric training toward a general practitioner-based model [3]. These changes contributed to a long-term shortage of pediatric specialists and reduced access to specialized care for children. In response to the growing workforce gap, Kazakhstan initiated the gradual reinstatement of pediatric training programs from 2017 to 2018 [4]. Nevertheless, despite the resumption of education, workforce capacity remains limited, particularly in highly specialized fields such as pediatric otorhinolaryngology (ear, nose, and throat, ENT) [5]. An additional constraining factor is the ongoing optimization of inpatient care, including reductions in hospital bed capacity and shorter lengths of stay [6], which may further limit the timely diagnosis and management of severe and complicated ENT conditions in children.

In this context, acute mastoiditis (AM) represents a useful clinical model for assessing the impact of systemic constraints within pediatric otorhinolaryngology. AM is a severe bacterial infection of the mastoid process that most commonly develops as a complication of acute otitis media and is characterized by inflammation and destruction of mastoid air cells, with a risk of infection spreading to adjacent anatomical structures [7,8]. Although the incidence of AM has declined in high-income countries, it remains a clinically important condition, particularly in pediatric practice [9]. According to published data, the global incidence of mastoiditis ranges from 1.2 to 1.6 cases per 100,000 people per year, with the highest burden observed among young children [10,11]. The increased vulnerability of this age group is attributed to immune system immaturity [12], as well as the increasing prevalence of antimicrobial resistance [13,14]. The reported rates of intracranial complications associated with AM vary from 5% to 29% and include sigmoid sinus thrombosis, epidural and subdural abscesses, suppurative labyrinthitis, and facial nerve paralysis [15].

Despite its clinical importance, systematically collected national-level epidemiological data on the incidence of acute otitis media and its complications, including AM, are largely lacking in Kazakhstan. The available evidence remains fragmented and is typically confined to reports from individual hospitals or referral centers, as not all healthcare facilities have the capacity to provide specialized ENT care. Another restriction is the absence of an updated national clinical guideline [16] that provides clearly defined management strategies for complicated forms of otitis media and mastoiditis, which may contribute to variability in clinical decision-making and delays in timely referral to specialized institutions.

Accordingly, the aim of the present study was to assess and forecast healthcare system indicators in Kazakhstan via AM as a model condition while simultaneously examining the clinical and epidemiological characteristics of AM.

2. Materials and Methods

2.1. Study Design

This study was conducted in several two stages. This integrated, two-part design was implemented to address the management of pediatric AM from a systems-based perspective. First, national healthcare statistics were analyzed to forecast future population needs in Kazakhstan. This macrolevel analysis aimed to identify systemic trends, potential resource gaps, and the epidemiological context of pediatric ENT care at the national level, providing a backdrop for clinical findings. Second, a retrospective review of medical records from a large Kazakhstani tertiary referral center in Almaty was conducted to characterize the contemporary clinical presentation, management pathways, and outcomes of AM; data from the tertiary referral center in the capital city of Astana were not available for analysis. This single-center, in-depth clinical analysis aimed to provide detailed real-world insights into disease presentation and current management practices that are not routinely captured in national healthcare statistics.

2.2. Evaluation of Nationwide Demographic and Healthcare Data

The official healthcare statistics were obtained from the statistical yearbooks “Health of the Population of the Republic of Kazakhstan and Activities of Healthcare Organizations”, compiled by the Ministry of Healthcare of the Republic of Kazakhstan and publicly available on the website of the Salidat Kairbekova National Scientific Center for Healthcare Development [17]. The retrieved data included (i) the number of pediatricians per 10,000 people, (ii) the number of ENT doctors per 10,000 people, (iii) the availability of ENT hospital beds per 10,000 people, and (iv) the share of ENT surgeries within the overall volume of surgical procedures for ear surgeries. The healthcare statistics used in this study spanned the period from 1998 to 2024, which represents the full range of publicly accessible national data.

2.3. Retrospective Analysis of Pediatric AM Patients

A descriptive analysis was performed at a major referral center in Almaty, Kazakhstan (ENT Department, City Clinical Hospital No. 5). Medical records were extracted from the computerized hospital database and included all children admitted with AM between 1 September 2022, and 29 February 2024. Children were admitted to the center with a diagnosis of AM, which was confirmed by an otolaryngologist. The diagnosis was based on the presence of AOM, fever, otalgia, and retroauricular swelling and erythema. Medical records were reviewed for demographic data (age and sex), clinical presentations (fever, retroauricular erythema and swelling, otorrhea, pain on palpation, and fluctuance), laboratory parameters (hemoglobin, leukocyte count, platelet count, and ESR), and radiological findings (temporal bone computed tomography). Additionally, the type of complication (if present) and hospitalization details (total hospital stay and length of postoperative hospitalization in cases requiring surgery) were recorded. The inclusion criteria were as follows: (a) age 0–18 years and (b) the presence of clinical and radiological signs of AM [18]. Radiological confirmation via temporal bone computed tomography (CT) was required for all included patients. CT findings were used both to confirm the diagnosis of AM and to determine disease severity. Accordingly, incipient mastoiditis was classified as a moderate form of AM, whereas coalescent mastoiditis was defined as severe [19]. As

severity classification relies exclusively on radiological criteria, the observed distribution of moderate and severe cases reflects radiologically confirmed disease.

2.4. Ethics

The study was approved by the Ethics Committee of Kazakhstan's Medical University "Kazakhstani School of Public Health" (Approval No. 8, dated 6 May 2025). Parental informed consent was obtained for the use of retrospective medical records in accordance with institutional and national ethical requirements. All procedures were conducted in compliance with the principles of the Declaration of Helsinki.

2.5. Statistical Analysis

Healthcare statistics covering the period from 1998 to 2024 were collated in Microsoft Excel© for subsequent analysis. Historical trends were quantified via the average annual percentage change (AAPC), which summarizes the mean yearly change over the study period and was calculated from log-linear regression models fitted to the time series data. AAPC estimates are presented with corresponding 95% confidence intervals (CIs) to reflect statistical uncertainty. The forecasting of epidemiological trends through 2030 was performed via time series analysis in SPSS. The Expert Modeler function was applied to automatically identify the most appropriate forecasting model on the basis of goodness-of-fit statistics and residual diagnostics. Depending on the characteristics of the data, autoregressive integrated moving average (ARIMA) models were selected for series exhibiting autocorrelation and potential nonstationarity, whereas Holt's exponential smoothing models were used for series demonstrating a consistent linear trend without pronounced seasonality. Model adequacy was assessed via standard fit criteria and examination of residuals.

Projections are reported as point estimates with corresponding 95% confidence intervals. The width of the 95% confidence intervals was used to reflect uncertainty in the projections, with wider intervals indicating lower precision of long-term forecasts. Projected point estimates were therefore interpreted as indicative trends rather than exact future values, and all results are reported with corresponding confidence intervals to ensure transparent uncertainty assessment. Visualizations were generated to depict both observed and projected values.

Continuous variables are presented as medians with interquartile ranges (IQRs). The normality of continuous data was assessed via the Shapiro–Wilk test. As the data were nonnormally distributed, between-group comparisons for continuous measures were performed via the Mann–Whitney U test. Categorical variables are presented as counts and percentages and were compared between groups via the chi-square test. All the statistical analyses were conducted via SPSS for Windows, version 24.0 (IBM Corp., Armonk, NY, USA). A two-sided p value < 0.05 was considered statistically significant.

3. Results

3.1. Longitudinal Trends and Forecasted Availability of Pediatric and ENT Healthcare Resources

Table S1 presents longitudinal data on pediatric and otorhinolaryngology healthcare resources in Kazakhstan from 1998 to 2024 (Table S1). The number of pediatricians per 10,000 population showed considerable variability over the study period. After a peak of 5.5 in 2002, a general declining trend was observed, with values falling from 4.0 in 2011 to 2.5 in 2024. The AAPC was -2.58% (95% CI: $-3.16, -2.00$; $p < 0.001$), indicating a statistically significant annual decrease over the full period. The number of ENT doctors remained relatively low and stable, fluctuating between 0.1 and 1.0 per 10,000 people. From 2011 to 2024, the rate stabilized at 0.6–0.7. The AAPC of 1.05% was not statistically significant

(95% CI: $-1.28, 3.44$; $p = 0.183$). The availability of ENT hospital beds per 10,000 people exhibited a clear downward trajectory, declining from 1.0 in 1998 to 0.5 by 2024. The AAPC was -3.62% (95% CI: $-4.19, -3.05$; $p < 0.001$), reflecting a significant and steady reduction in bed capacity. The share of ENT surgeries among all surgical procedures demonstrated a consistent upward trend, rising from 0.4% in 1998 to 1.3% in 2024, with notable increases in 2008 and 2018–2024. The AAPC was 4.29% (95% CI: $3.59, 5.00$; $p < 0.001$), indicating a significant annual increase in the relative volume of ENT surgeries.

Table 1 presents the projected values for key pediatric and otorhinolaryngology healthcare indicators in Kazakhstan for the period 2027–2035, which are based on time series modeling of historical data. The projected number of pediatricians per 10,000 population is expected to remain stable at approximately 2.6–2.7 throughout the forecast period, with widening 95% confidence intervals over time. The ARIMA (4,1,0) model indicates a statistically significant declining trend in the historical data ($p < 0.001$). The number of ENT doctors is projected to remain unchanged at 0.6 per 10,000 population across all forecasted years, as supported by a significant ARIMA (0,0,1) model ($p < 0.001$). In contrast, the availability of ENT hospital beds per 10,000 people is projected to decrease gradually from 0.5 in 2027 to 0.3 by 2035. This downward trend, modeled via ARIMA (0,1,0), was marginally nonsignificant in the historical series ($p = 0.057$). The share of ENT surgeries within the total volume of surgical procedures is projected to increase steadily from 1.3% in 2027 to 1.5% by 2035. The Holt model did not demonstrate a statistically significant historical trend ($p = 0.468$), although a gradual increase is forecasted.

Table 1. The projected numbers of pediatricians, ENT doctors, ENT hospital beds, and shares of ear-related surgeries in Kazakhstan for the years 2026 and 2030, accompanied by 95% confidence intervals.

Year	Number of Pediatricians (per 10,000 Population) Number (95% CI)	Number of ENT Doctors (per 10,000 Population) Number (95% CI)	Availability of ENT Hospital Beds per 10,000 Population Number (95% CI)	Share of ENT Surgeries within the Overall Volume of Surgical Procedures Ear Surgeries Number (95% CI)
2027	2.6 (1.6; 3.7)	0.6 (0.2; 1.1)	0.5 (0.3; 0.6)	1.3 (1.1; 1.5)
2028	2.6 (1.3; 3.9)	0.6 (0.2; 1.1)	0.4 (0.3; 0.6)	1.3 (1.1; 1.5)
2029	2.7 (1.2; 4.2)	0.6 (0.2; 1.1)	0.4 (0.2; 0.6)	1.3 (1.1; 1.6)
2030	2.6 (1.1; 4.2)	0.6 (0.2; 1.1)	0.4 (0.2; 0.6)	1.4 (1.2; 1.6)
2031	2.6 (1.1; 4.2)	0.6 (0.2; 1.1)	0.4 (0.1; 0.6)	1.4 (1.2; 1.6)
2032	2.6 (1.0; 4.2)	0.6 (0.2; 1.1)	0.4 (0.1; 0.6)	1.4 (1.2; 1.7)
2033	2.6 (1.0; 4.2)	0.6 (0.2; 1.1)	0.3 (0.1; 0.6)	1.5 (1.3; 1.7)
2034	2.6 (0.9; 4.3)	0.6 (0.2; 1.1)	0.3 (0.0; 0.6)	1.5 (1.3; 1.7)
2035	2.6 (0.8; 4.4)	0.6 (0.2; 1.1)	0.3 (0.0; 0.6)	1.5 (1.3; 1.7)
Model parameters	ARIMA (4, 1, 0), $p < 0.001$	ARIMA (0, 0, 1), $p < 0.001$	ARIMA (0, 1, 0), $p = 0.057$	Holt $p = 0.468$

Figure 1 illustrates the forecasted availability of key pediatric and otorhinolaryngology healthcare indicators in Kazakhstan for the period 2026–2030 and complements the numerical projections presented in Table 1. The number of pediatricians per 10,000 population is projected to remain largely stable over the forecast horizon (Figure 1A). Similarly, the number of ENT specialists is expected to show minimal change, maintaining a constant level throughout the period (Figure 1B). In contrast, the availability of ENT hospital beds

demonstrated a slight downward trend (Figure 1C). Conversely, the share of ENT surgeries within the overall volume of surgical procedures is projected to increase gradually over time (Figure 1D), indicating a growing surgical workload despite a stable workforce capacity.

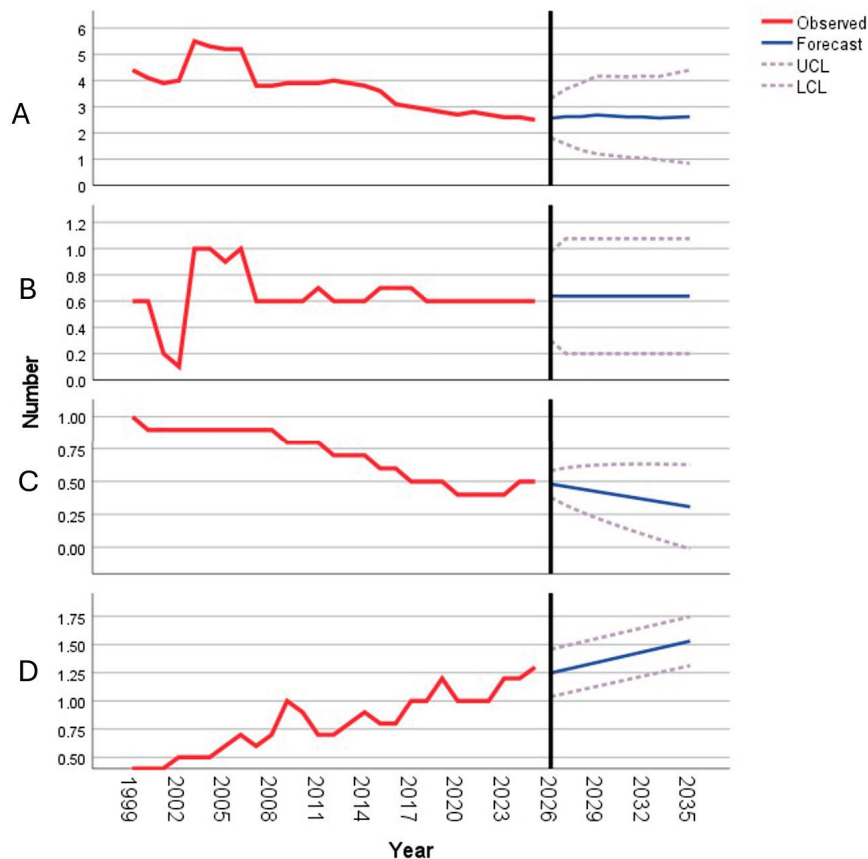


Figure 1. Forecasted availability of pediatricians (A), ENT specialists (B), ENT hospital beds (C), and ENT surgeries (D) in Kazakhstan (2026–2030). (UCL—Upper Confidence Limit; LCL—Lower Confidence Limit).

3.2. Comparison of the Clinical Characteristics of Noncomplicated and Complicated Acute Mastoiditis Patients

To contextualize these national healthcare resource trends within patient outcomes, a clinical case series of pediatric acute mastoiditis patients was analyzed, and the results are presented below. Over an 18-month retrospective observation period, 95 patients with AM who met the inclusion criteria were identified. The mean age of the patients was 74.92 months, with a range from 4 to 193 months. Of these, 58.9% ($n = 56$) were male, and the vast majority resided in urban areas. All 95 patients presented with fever, swelling, otorrhea, and pain on palpation. Retroauricular erythema was observed in 94.7% of the patients ($n = 90$), whereas fluctuations were detected in 43.2% of the patients ($n = 46$). In 42.1% of the patients ($n = 40$), AM affected the left side; right-sided involvement was observed in 35.8% ($n = 34$), and bilateral AM was identified in 22.1% of the patients ($n = 22$). Eighty-nine patients were admitted to the hospital with moderate disease severity, while six were classified as severely ill. All patients had adenoiditis as a comorbidity.

Complications developed in 38 patients with AM (40%). Among the observed complications, subperiosteal abscess was identified as one of the most common manifestations ($n = 20$), followed by zygomatic abscess ($n = 13$). Isolated cases of sigmoid sinus thrombosis ($n = 1$), facial paresis ($n = 1$), a combination of facial paresis and meningitis ($n = 1$), sub-

periosteal abscess with sigmoid sinus thrombosis ($n = 1$), and subperiosteal abscess with facial paresis ($n = 1$) were also identified.

Table 2 presents the differences between patient subgroups with noncomplicated AM and those with complicated AM. The median age and sex ratio did not differ significantly between the groups ($p = 0.882$ and $p = 0.055$, respectively). Urban residency was prevalent in both groups, with no significant difference ($p = 0.449$). Severe cases were observed exclusively in the complicated AM group (15.8%, $p = 0.002$), as was rapid disease progression within 7 days (7.9%, $p = 0.031$). Retroauricular erythema was common in both groups, although it was more common in complicated cases (100% vs. 91.2%, $p = 0.061$), whereas retroauricular fluctuance was significantly different (76.3% vs. 21.1%, $p = 0.001$). Radiologically, temporal bone destruction on CT was markedly more prevalent in the complicated group (84.2% vs. 3.5%, $p = 0.001$). Laboratory parameters revealed a significantly greater leukocyte count ($p = 0.001$) and ESR ($p = 0.019$) in complicated patients than in uncomplicated patients, whereas hemoglobin and platelet counts were not significantly different. Patients with complicated AM had longer total hospital stays (median of 5 vs. 4 days, $p = 0.001$) and longer postoperative hospitalization periods (median of 4 vs. 3 days, $p = 0.001$).

Table 2. Comparative characteristics of patients with noncomplicated and complicated AM.

	Noncomplicated AM ($n = 57$)	Complicated AM ($n = 38$)	<i>p</i> Value
Age (Me (range)) (months)	63 (33–106)	70 (34–90)	0.882
Male/Female Ratio	1.66	1.24	0.055
Urban residency	51 (89.5%)	32 (84.2%)	0.449
Clinical presentation			
Severe cases	0 (0%)	6 (15.8%)	0.002
Rapid progress (<7 days)	0 (0.0%)	3(7.9%)	0.031
Retroauricular erythema	52 (91.2%)	38 (100.0%)	0.061
Retroauricular fluctuance	12 (21.1%)	29 (76.3%)	0.001
CT findings			
Destruction of temporal bone	2 (3.5%)	32 (84.2%)	0.001
Laboratory parameters			
Hemoglobin (Me (range)) (g/L)	113 (104–121)	115 (104–121)	0.948
Leucocyte count (Me (range)) ($\times 10^9$ /L)	10 (9–13)	15 (11–17)	0.001
Platelet count (Me (range)) ($\times 10^9$ /L)	344 (306–458)	346 (252–470)	0.477
ESR (Me (range)) (mm/h)	37 (27–48)	42 (38–49)	0.019
Hospitalization details			
Total hospital stay (days)	4 (4–4)	5 (5–5)	0.001
Length of postoperative hospitalization (in case of surgery) (days)	3 (3–3)	4 (4–4)	0.001

Table 3 shows the distribution of surgical interventions among patients diagnosed with noncomplicated and complicated AM. The most frequently performed procedure among patients with noncomplicated AM ($n = 57$) was myringotomy, reported in 96.5% of patients (55/57), whereas it was performed in only 15.8% (6/38) of patients with complicated AM ($n = 38$). Conversely, atticotomy was predominant in the complicated AM group,

with a frequency of 60.5% (23/38), compared with only 1.8% (1/57) in the noncomplicated group. A combination of myringostomy on the contralateral side with atticoantrotomy was performed in 13.2% (5/38) of the complicated patients and in 1.8% (1/57) of the noncomplicated patients. Mastoidectomy was exclusively reported in the complicated group, accounting for 10.5% (4/38) of the cases, and was not performed in any of the noncomplicated patients.

Table 3. Types of surgeries performed in patients with noncomplicated and complicated AM.

Type of Intervention	Noncomplicated AM (n = 57)	Complicated AM (n = 38)
Myringostomy	55 (96.5%)	6 (15.8%)
Atticoantrotomy	1 (1.8%)	23 (60.5%)
Myringostomy on the other side in combination with atticoantrotomy	1 (1.8%)	5 (13.2%)
Mastoidectomy	0 (0.0%)	4 (10.5%)

4. Discussion

4.1. Forecasted Healthcare Demand and Workforce Capacity Challenges

A negative AAPC indicates a steady decrease in pediatrician availability in Kazakhstan, which is consistent with the long-term consequences of earlier healthcare system reforms. Analytical reports by the World Health Organization (WHO) indicate that such workforce shifts may increase the risk of delayed detection of severe conditions in children and contribute to a greater proportion of complicated cases requiring inpatient care and surgical intervention [3]. In this context, assessments of pediatric care quality in hospitals in Kyrgyzstan and Tajikistan have shown that shortages of pediatricians substantially limit the capacity of healthcare facilities to deliver high-quality medical services [20]. Moreover, Sakai, R. et al. (2016) demonstrated that higher pediatric workforce density is associated with lower mortality rates among children under five years of age [21].

In light of this, a persistently rising percentage of ENT surgical procedures and a comparatively stable but low density of otorhinolaryngologists point to the establishment of a mismatch between the supply and demand of specialized treatment. A similar “supply–demand gap” has been described in the international literature on workforce planning in otorhinolaryngology, where modeling studies demonstrate the risk of misalignment between population needs and specialist availability when service demand increases more rapidly than workforce capacity [22,23]. In Kazakhstan, this imbalance may be further exacerbated by the continued growth of the pediatric population [24], leading to increased utilization of ENT services and an increasing need for surgical care.

In addition, reductions in inpatient bed capacity occurring alongside increasing surgical volumes and continued growth of the pediatric population may constrain the ability of healthcare systems to adequately manage severe and complicated conditions, including AM. International experience indicates that the optimization of inpatient care without concurrent strengthening of outpatient and specialized services is associated with extended hospital stays, increased pressure on surgical departments, and a heightened risk of delays in the delivery of timely care for acute pediatric conditions [25].

4.2. Incidence and Complication Profile of Pediatric AM

Over an 18-month retrospective observation period at a referral center in Almaty, Kazakhstan, 95 pediatric patients were diagnosed with AM, indicating an exceptionally high case burden over a relatively short timeframe. This finding is particularly notable

compared with international reports [26–28]. For example, a review of electronic medical records from one of the largest pediatric hospitals in the United States identified only 129 AM cases over an eight-year period (2011–2019) [26], whereas Karaaslan et al. (2023) reported only 28 hospitalizations for AM over seven years at a municipal hospital in Istanbul, Turkey [27]. In contrast, data from the Israeli national registry revealed an incidence of 7.78 cases per 100,000 child-years, exceeding rates reported in other Western countries [28].

The 40% AM complication rate reported in this study is higher than that reported in several investigations worldwide [29]. This difference is likely multifactorial and may partly reflect referral bias, as the clinical cohort was drawn from a tertiary referral center that predominantly manages more severe and advanced cases. In addition, diagnostic thresholds and case definitions, including the routine use of CT, may have contributed to a higher detection rate of bony involvement and complications than studies using more conservative diagnostic approaches. Furthermore, the high proportion of complicated cases may reflect delayed outpatient diagnosis and late hospital presentation, as indicated by the frequent presence of retroauricular erythema and fluctuation and radiological evidence of bone destruction in 84.2% of patients.

A comparison with published data revealed substantial variability in the reported complication rates of AM, ranging from 1.9% to 35% [29]. For example, Guillén-Lozada et al. (2022) reported that 109 of 412 children with AM (26.5%) developed severe intracranial complications, which significantly influenced treatment strategies and the length of hospital stay [30]. In contrast, another study reported complications in only 9.5% of 147 AM patients and reported a significant inverse association between retroauricular erythema and the likelihood of complications [29].

In the present study, elevated leukocyte counts and ESR emerged as the most informative laboratory markers in complicated AM, underscoring the importance of close laboratory monitoring to prevent adverse outcomes. These findings are supported by a study from Lisbon involving 135 children with AM, which demonstrated a significant association between increased leukocyte counts, elevated C-reactive protein levels, and the development of complications [31]. Moreover, several authors emphasize the ESR as a particularly valuable parameter for guiding decisions regarding both antimicrobial therapy and the need for surgical intervention [32].

Myringotomy was the predominant surgical intervention in uncomplicated AMs (96.5%), whereas atticoantrotomy was more common in complicated cases (60.5%), reflecting disease severity. This pattern aligns with published data, as myringotomy is a minimally invasive procedure used for middle ear drainage, whereas atticoantrotomy and, less frequently, mastoidectomy (10.5%) are reserved for advanced cases with bony involvement [33,34]. Some authors, including Mierzwiński et al. (2019), have suggested early mastoidectomy in patients unresponsive to 48 h of intravenous antibiotic therapy to reduce the risk of recurrence [35].

4.3. System-Level Preventive Strategies and Policy Implications

Kazakhstan's healthcare system continues to face structural challenges, including workforce shortages, medical staff migration, and pronounced urban–rural disparities in resource distribution [36]. Additional constraints arise from the high level of infrastructure depreciation and the misalignment of healthcare tariffs with actual service costs and inflationary pressures [37]. Against this background, the clinical profile of pediatric AM observed in the present study—characterized by a high proportion of complicated cases—may reflect the downstream effects of workforce shortages, uneven access to timely care, and limitations at the primary and secondary care levels. However, preventing complicated

forms of pediatric diseases cannot be viewed solely through the lens of increasing physician numbers and instead requires coordinated, system-level interventions.

One key priority is maintaining high immunization coverage, as declining vaccination rates against vaccine-preventable infections have been associated with an increased incidence of otitis media and related conditions [38]. For example, Daniel et al. (2013) demonstrated that routine pneumococcal vaccination was associated with a significant reduction in invasive ENT infections and pediatric hospitalizations [39]. Equally important is improving parental health literacy and awareness, given the persistent trends of vaccine refusal and self-medication among caregivers in the country [40].

Systematic strengthening of healthcare workforce capacity, including continuous professional development supported by remote education and telemedicine tools [41], as well as sustained interdisciplinary collaboration between clinicians and microbiological laboratories, may improve early diagnosis and optimize treatment strategies. At the same time, the absence of a unified, evidence-based clinical management pathway for complicated pediatric infections highlights the need for the development of standardized approaches aligned with international guidelines and adapted to local healthcare system capacities [16]. According to the Organization for Economic Co-operation and Development (OECD), strengthening research capacity in primary health care and expanding the clinical and decision-making roles of general practitioners represent some of the most effective approaches to reducing pressure on inpatient and specialized care services [42]. Finally, revising remuneration policies, implementing targeted retention and incentive strategies for physicians in both urban and rural settings, developing dedicated investment programs for healthcare infrastructure modernization are essential to ensure the long-term sustainability and resilience of the national healthcare system.

4.4. Study Limitations

This study has several limitations that should be acknowledged. First, the retrospective design based on medical record review may be subject to information bias, as the completeness and quality of documentation could not be fully controlled. Although all available clinical, laboratory, and radiological data were extracted, variations in record-keeping practices may have influenced the consistency of certain variables.

Second, the clinical analysis was conducted at a single tertiary referral center, which may limit the generalizability of the findings. Referral bias may have contributed to the higher observed incidence and complication rates of AM, and the results may not fully represent national epidemiological trends. Moreover, reliance on AM as a sole indicator may limit the robustness of conclusions regarding healthcare system constraints, which are ideally evaluated via multiple complementary conditions.

Third, the absence of a centralized national registry for acute otitis media and its complications in Kazakhstan restricts the ability to validate findings against population-based data.

5. Conclusions

AM should not be regarded as a direct measure of healthcare system performance but may serve as a contextual indicator reflecting pressures within specific components of the healthcare system, particularly specialized and inpatient services. The observed trends suggest that system-level constraints may influence clinical presentation and management; however, broader validation across multiple centers, levels of care, and clinical conditions is needed to confirm the generalizability of these findings. Overall, this study highlights the value of integrating health system indicators with clinical data to better characterize emerging challenges in pediatric care and underscores the need for future studies incorporating

additional sentinel conditions to assess healthcare system capacity more comprehensively and support evidence-based planning and policy development.

Supplementary Materials: The following supporting information can be downloaded at: <https://www.mdpi.com/article/10.3390/children13020170/s1>. Table S1: The numbers of pediatricians, ENT doctors, ENT hospital beds, and shares of ear-related surgeries in Kazakhstan (1998–2024).

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Abbreviations

The following abbreviations are used in this manuscript:

AM	Acute mastoiditis
ESR	Erythrocyte sedimentation rate
CRP	C-reactive protein
CT	Computed tomography
MRI	Magnetic resonance imaging
RK	Republic of Kazakhstan
WHO	World Health Organization
AAPC	Average annual percentage change
ENT	Ear, nose, and throat
ARIMA	Autoregressive integrated moving average
CIs	Confidence intervals
IQRs	Interquartile ranges

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