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CONDUCTIVE HEARING LOSS WITHIN UNIVERSAL NEWBORN HEARING SCREENING PROGRAMS: A SYSTEMATIC REVIEW

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

Abstract

Background: Universal Newborn Hearing Screening (UNHS) attempts to identify children with a permanent, bilateral, moderate or greater hearing loss at birth. However, children who are referred from UNHS programs may have conductive hearing loss (CHL), sensorineural, or mixed hearing loss. The aim of this review was to investigate the prevalence, sub-classifications, audiological diagnosis, and medical management of CHL within UNHS programs.

Material and methods: A systematic literature search was completed in the scientific databases PubMed, CINAHL, and Embase. Studies were reviewed with reference to the inclusion criteria, then graded to assess the internal and external validity, leaving 25 studies for review.

Results: The prevalence of conductive hearing loss ranged from 0.4% to 64.5%. 'Genetic' and 'Permanent' were the only two sub-classifications of CHL identified, with no uniform terminology evident. Given CHL is not a target condition of UNHS, audiological assessment was consistent with the diagnosis of Permanent Childhood Hearing Loss (PCHL). There was little evidence of audiological review, onward referrals, or medical management for CHL within UNHS programs. Of the evidence obtained, no alternative pathway was found for children identified with CHL through UNHS.

Conclusions: In view of the limited evidence for CHL within UNHS, further investigation into the prevalence, sub-classification, and appropriate management of CHL within a UNHS program is recommended to better guide evidence-based assessment and management of these children.

Key words: audiological assessment • children • conductive hearing loss • infants • neonates • prevalence • universal newborn hearing screening

LA PÉRDIDA AUDITIVA DE CONDUCCIÓN EN EL PROGRAMA DE CRIBADO AUDITIVO UNIVERSAL EN RECIÉN NACIDOS: UNA REVISIÓN SISTEMÁTICA

Resumen

Introducción: El cribado auditivo universal neonatal (UNHS) está diseñado para identificar a niños con pérdida auditiva bilateral permamente de nivel moderado a grave en el momento de nacer. Sin embargo, los niños usuarios de los programas de UNHS pueden tener una pérdida auditiva de conducción (CHL), sensorial o mixta. El propósito de esta revisión fue investigar la incidencia, subclasificación, diagnóstico audiológico y manejo médico de CHL bajo los programas de UNHS.

Material y métodos: se realizó una revisión sistemática de la literatura en las bases de datos científicas PubMed, CINAHL y Embase. Los resultados obtenidos se revisaron en relación a los criterios de inclusión, y luego se evaluó su valor interno y externo, obteniendo así 25 trabajos adecuados para la revisión.

Resultados: La incidencia de pérdida auditiva conductiva varió de 0.4% a 64.5%. Las dos únicas subclasificaciones identificadas para CHL fueron perdidas auditivas "genéticas" y "permanentes" - no se utilizó una terminología uniforme. Teniendo en cuenta que CHL no es una condición objetivo detectada bajo UNHS, la evaluación audiológica estaba en línea con la evaluación diagnóstica de la pérdida auditiva permanente infantil (PCHL). Hubo poca evidencia de más pruebas audiológicas, derivación y tratamiento médico para CHL bajo los programas de UNHS. Los resultados de UNHS no permitieron encontrar una ruta alternativa para los niños con CHL.

Conclusiones: debido a la evidencia limitada de la existencia de CHL bajo UNHS, se recomienda una mayor investigación sobre la incidencia, subclasificación y conducta apropiada de CHL bajo el programa UNHS para evaluar los resultados basados en evidencia y gestionar mejor estos niños.

Palabras clave: evaluación audiológica • niños • pérdida auditiva conductiva • bebés • recién nacidos • prevalencia • cribado auditivo universal en recién nacidos

КОНДУКТИВНАЯ ТУГОУХОСТЬ В ПРОГРАММЕ УНИВЕРСАЛЬНОГО СКРИНИНГА СЛУХА НОВОРОЖДЕННЫХ: СИСТЕМАТИЧЕСКИЙ ОБЗОР

Аннотация

Введение: Целью универсального аудиологического скрининга новорожденных (UNHS) является выявление у детей постоянной двусторонней тугоухости средней или глубокой степени при рождении. Однако, дети, получившие направление в рамках программы UNHS, могут иметь кондуктивную (CHL), нейросенсорную или смешанную тугоухость. Цель данного обзора состояла в том, чтобы исследовать распространенность, подклассификации, аудиологическую диагностику и медицинскую процедуру в случае CHL в рамках программ UNHS.

Материалы и методы: В научных базах данных PubMed, CINAHL и Embase был проведен систематический обзор литературы. Полученные результаты были рассмотрены согласно критериям включения, а затем была проведена оценка внутренней и внешней достоверности, что в результате дало 25 исследований, подходящих для осуществления обзора.

Результаты: Распространенность кондуктивной тугоухости варьировалась от 0,4% до 64,5%. Было выявлено две подклассификации CHL: «генетические» и «постоянные». Не определено единой терминологии. Учитывая, что CHL не является целью проведения UNHS, аудиологическая оценка соответствовала диагнозу «Постоянная потеря слуха в детстве» (PCHL). Существует мало доказательств того, что в рамках программ UNHS при выявлении CHL проводилось аудиологическое наблюдение, дальнейшее направление на консультацию к специалисту или другие медицинские процедуры. Полученные результаты показывают, что не определено альтернативного пути для детей, с выявленнной кондуктивной тугоухостью в рамках программы UNHS.

Выводы: Ввиду ограниченности данных выявления кондуктивной тугоухости в рамках универсального аудиологического скрининга новорожденных, рекомендуется дальнейшее исследование распространенности, подклассификации и надлежащей медицинской процедуры при выявлении CHL в рамках программы UNHS для более точной оценки данного нарушения на основании соответствующих результатов исследований и опеки за детьми с данным типом заболевания.

Ключевые слова: аудиологическая оценка • дети • кондуктивная тугоухость •младенцы • новорожденные • распространенность • универсальный аудиологический скрининг новорожденных

NIEDOSŁUCH PRZEWODZENIOWY W PROGRAMIE UNIWERSALNYCH BADAŃ PRZESIEWOWYCH SŁUCHU U NOWORODKÓW: PRZEGLĄD SYSTEMATYCZNY

Streszczenie

Wstęp: Uniwersalne przesiewowe badania słuchu u noworodków (UNHS) mają na celu wykrycie u dzieci w chwili urodzenia trwałego obustronnego niedosłuchu w stopniu umiarkowanym lub wyższym. Jednak dzieci skierowane z programów UNHS mogą mieć niedosłuch przewodzeniowy (CHL), odbiorczy lub mieszany. Celem niniejszego przeglądu było zbadanie występowania, podklasyfikacji, diagnozy audiologicznej i postępowania medycznego w przypadku CHL w ramach programów UNHS.

Materiał i metody: Przeprowadzono systematyczny przegląd literatury w naukowych bazach danych PubMed, CINAHL i Embase. Uzyskane wyniki poddano przeglądowi w odniesieniu do kryteriów włączenia, a następnie oceniono ich wartość wewnętrzną i zewnętrzną, uzyskując w ten sposób 25 prac nadających się do przeglądu.

Wyniki: Częstość występowania niedosłuchu przewodzeniowego wynosiła od 0,4% do 64,5%. Jedynymi dwiema zidentyfikowanymi podklasyfikacjami dla CHL były niedosłuchy „genetyczne" i „trwałe" – nie stwierdzono stosowania ujednoliconej terminologii. Biorąc pod uwagę, że CHL nie jest docelowym schorzeniem wykrywanym w ramach UNHS, ocena audiologiczna była zgodna z oceną diagnostyczną trwałej utraty słuchu w dzieciństwie (PCHL). Było niewiele dowodów, że pacjentom z przewodzeniowym ubytkiem słuchu w ramach programów UNHS wykonano badania audiologiczne, następnie wydano skierowania czy przeprowadzono postępowanie medyczne. Wyniki uzyskane w ramach UNHS nie pozwoliły na znalezienie alternatywnej ścieżki dla dzieci, u których wykryto CHL.

Wnioski: W związku z ograniczonymi dowodami na wykrywanie CHL w ramach UNHS zaleca się dalsze badania w sprawie występowania, podklasyfikacji oraz odpowiedniego postępowania w przypadku CHL w ramach programu UNHS, aby lepiej oceniać wyniki na podstawie dowodów i radzić sobie z dziećmi z niedołuchem przewodzeniowym.

Słowa kluczowe: ocena audiologiczna • dzieci • niedosłuch przewodzeniowy • niemowlęta • noworodki • rozpowszechnienie • uniwersalne badania przesiewowe słuchu u noworodków

Abbreviations

UNHS – universal newborn hearing screening
CHL – conductive hearing loss
PCHL – permanent childhood hearing loss
OM – otitis media
VRA – visual response audiometry
PTA – pure tone audiometry
TEOAE – transient evoked otoacoustic emission
DPOAE – distortion product otoacoustic emission
GP – general practitioner
ENT – ear nose and throat
ANSD – auditory neuropathy spectrum disorder
USPSTF – US Preventive Services Task Force
aABR – automated auditory brainstem response
ABR – auditory brainstem response
ASSR – auditory steady-state response

Background

Newborn hearing screening has been implemented throughout the world, leading to significant advances in early identification of children with hearing loss. The process includes screening, diagnostic audiology assessment, and

onward management pathways. The aim of Universal Newborn Hearing Screening (UNHS) is to identify infants and children with the target condition of bilateral moderate, or greater, permanent childhood hearing loss (PCHL). Historically, support for this target condition was established through evidence linking the provision of early intervention for these children with improved speech and language outcomes [1]. As this criterion is uniform across all UNHS programs, significant research has been dedicated to finding the optimal assessment pathways for these children [1–8]. However, through the same hearing screening process, many infants and children have also been identified with a hearing loss that does not fall into the target category. These non-target hearing conditions include: minimal or mild permanent hearing loss, unilateral permanent hearing loss, and conductive hearing loss (CHL). While the literature shows that these children are also at risk of social, academic, and speech and language difficulties [9–11], there is little evidence to guide the identification, assessment, and management of these children within UNHS programs. Unfortunately, existing pathways for children with PCHL are often not appropriate for children identified with a CHL.

A major cause of CHL in children is otitis media (OM). OM is highly prevalent in paediatric populations and refers to a group of inflammatory diseases of the middle ear space, often occurring alongside a range of bacterial infections in the upper respiratory tract [12–14]. Research indicates that long-standing or chronic CHL that is present during critical periods of language development places children at risk of speech and language delay and anxiety and depression disorders, leading to poorer social, educational, and vocational outcomes [15–17]. UNHS offers a unique opportunity for early identification of CHL. However, appropriate assessment and interventions for CHL within UNHS programs have yet to be addressed.

CHL can be attributed to either congenital or acquired aetiologies [18]. Acquired CHL can be the result of many causes, including OM, excessive cerumen, foreign bodies, and cholesteatoma [18–20]. Several congenital factors are also associated with CHL, often resulting from deficits in the development of the ear while in utero, as in children born with microtia or atresia [19]. Similarly, craniofacial anomalies such as cleft palate and cleft lip are often linked to middle ear anomalies that typically result in CHL [18]. There is also a clear association between a number of syndromes present at birth and the occurrence of CHL throughout childhood [21–22].

Given the number of aetiologies resulting in CHL, it is not surprising the CHL in infants and children is commonly identified through UNHS programs. Studies have reported that up to 11% of infants referred on UNHS have a CHL [23], and this rate is often higher than children diagnosed with PCHL [24–25]. Despite its prevalence within UNHS, CHL has largely been considered a false positive on UNHS, often associated with excessive appointments and over-testing, resulting in undue stress on both the parents or guardians and children [24,26–27].

While CHL is regularly identified through UNHS, few screening programs have categorised this type of hearing loss to guide early intervention. Some programs classify CHL as permanent if the hearing loss cannot be attributed to non-structural, middle ear temporary conditions, such as OM [28]. The term 'Genetic CHL' has been used if the hearing loss is associated with a syndrome strongly linked with CHL, craniofacial anomalies, or causes other than OM [29]. Despite these terminologies, there has been little guidance over optimal intervention pahways for these children. Overall, universal evidence-based sub-classifications of CHL are absent within UNHS programs. Establishment of sub-classifications of CHL to guide the assessment and management of these children might potentially reduce developmental delays for high-risk children.

Accurate audiological assessment of middle ear conditions and of CHL within paediatric populations has been a challenge, with most research evaluating the efficacy of tympanometric measures using a variety of probe tones across age groups [30–33]. More recently, the application of wide-band absorbance has grown in popularity as a tool for the assessment of middle ear dysfunction and associated hearing loss. While its application is not yet standard practice, research has demonstrated improved accuracy in the assessment of CHL in older children in comparison to traditional tympanometry (at 226 Hz), with promising results in the diagnosis of CHL in infants [32,34]. Nevertheless, a battery of tests is often recommended to determine the type and degree of hearing loss in paediatric populations [35]. A test battery can include a conditioned behavioural response such as visual response audiometry (VRA) (6–24 months) or play audiometry (3–8 years), as well as otoacoustic emissions (including transient evoked otoacoustic emissions (TEOAEs) and distortion product otoacoustic emissions (DPOAEs)) and auditory evoked potentials [36]. Once CHL is identified, regular monitoring of hearing can occur along with referral for hearing amplification or medical management if required [11]. While some management pathways for CHL within UNHS exist, evaluation of these management pathways has received little attention in comparision with PCHL.

Given CHL is not the focus of UNHS programs, few evidence-based guidelines exist to direct audiological diagnostic assessment or management of CHL identified through such a program. While some screening programs recommend a hearing review at 12 months of age [37], others recommend a review at 4–8 weeks followed by a referral to a general practitioner (GP) or ear nose and throat specialist (ENT) if the condition persists [38]. Referral for hearing amplification is often only discussed in instances of likely chronic or long-standing CHL, such as children with cleft palate or Down syndrome [28,38]. Frequently, children with CHL are discharged from screening programs. As such, they are more likely to be lost to follow-up, be later diagnosed with a hearing loss, and are less likely to be fit for hearing amplification at an opportune time [4,28,39]. These findings show variation in the ongoing management for CHL and highlight the need for the development of a protocol for the on-going assessment and management of CHL within a UNHS.

Medical management for CHL typically involves a review by a GP and referral to an ENT specialist if chronic occurrence is indicated [40]. One management option is surgery, where fluid is removed from the middle ear

space through myringotomy with or without tympanostomy tube insertion. The efficacy of myringotomy in the absence of tympanostomy tube insertion has been questioned, with more favourable outcomes for tympanostomy tube insertion for management of middle ear fluid [41,42]. There have also been mixed results regarding the benefits of tympanostomy tube insertion on long-term hearing outcomes and speech development [43–45]. Despite these findings, the relationship between earlier identification of CHL within UNHS and a review of hearing outcomes following medical management of CHL have yet to be addressed in the literature.

The aim of this review was to thoroughly investigate the prevalence and sub-classifications of CHL within UNHS programs, including current audiological and medical management for children identified through a newborn hearing screen. In addressing the aim of the review, the following research questions were developed:

- What is the prevalence of CHL within UNHS programs?
- Are there any sub-classifications of CHL applied within UNHS programs?
- How is CHL assessed and what onward referrals are made within UNHS programs?
- What is the current medical management of children identified with a CHL within UNHS programs?

Material and methods

To ensure sensitivity to the research purpose and that the study selection was systematic and impartial, inclusion and exclusion criteria were developed [46–47]. A search strategy was then developed, including selection of scientific databases and search terms. Next, appraisal of the literature was conducted to evaluate the applicability of the papers to the review questions and the overall methodological quality. Finally, detailed data analysis was conducted from the included studies to summarise the current literature and answer the research questions.

Types of studies

Inclusion criteria
- Empirical, qualitative, quantitative, and cohort studies.

Exclusion criteria
- Case studies, purely theoretical publications, grey matter (media, commentaries, etc.), and studies where English translation could not be sourced.

Types of participants

Inclusion criteria
- Infants and children referred from UNHS for audiology assessment irrespective of referral type, screening methodology, or place in the health care pathway.
- Children up to 16 years of age at the time of assessment who had been referred from a newborn hearing screening program.

Exclusion criteria
- Infants and children who were not referred for audiology assessment from a UNHS program.
- Infants and children identified with a permanent hearing loss from a universal newborn hearing screening program, including auditory neuropathy spectrum disorder (ANSD) and retrocochlear disorder.
- Infants and children identified with a mixed hearing loss were excluded as the health care pathway would likely differ from that of a CHL.
- Case studies and studies examining animal subjects.
- Infants and children seen through hearing screening programs where referral was not initiated from a UNHS program.

Types of outcome measures

Prevalence was defined as the frequency of CHL within a population in both raw and descriptive form. Where whole numbers were available, but descriptive statistics not reported, the frequency was calculated to allow comparison between studies. Audiological and medical management was defined as any referral or onward process with the primary aim of assessing or improving OM or CHL.

Search strategy and retrieval process

The literature search was conducted in October 2015 using the peer-reviewed electronic databases of PubMed, CINAHL, and Embase. A repeat search was completed on 8 March, 2018.

The search utilised a list of key words which were then customised to the search protocols unique to each database. MeSH terms were then applied to provide a comprehensive result. This search yielded words with reference to CHL (that is, *transient hearing loss*), prevalence (*incidence, proportion, frequency*), assessment (*investigation*, *description*, *characterization*, *characterisation*, and *intervention*), management (*treatment*, *therapy*, *surgery*, *remedy*, or *test*), and newborn hearing screening programs (*targeting surveillance* and *targeted surveillance programs*). From these searches, the titles and abstracts were assessed using the inclusion and exclusion criteria. Finally, review of the reference lists of the selected studies was conducted.

Quality assessment

Methodological analysis was conducted in three stages to assess internal and external validity. First, an overall measure of evidence strength was determined to yield high quality studies with minimum risk of errors [48]. The criteria developed by the U.S. Preventative Services Task Force (USPSTF) were deemed appropriate due its previous application in large-scale preventative health care studies [49–50]. All studies were excluded at the lowest level of evidence (level III). An adaptation of the McMaster grading tool was used to assess the internal validity of the publications, due to its appraisal of both qualitative and quantitative studies and its application in reviews of comparable health care programs [50–52]. This tool evaluated each study on several elements including study purpose, review of the literature, study design, data collection, analysis, and overall outcomes and conclusions. To be included, the study needed an overall score of greater than 5. A measure of external validity was included to ensure that the evidence obtained could be applied within

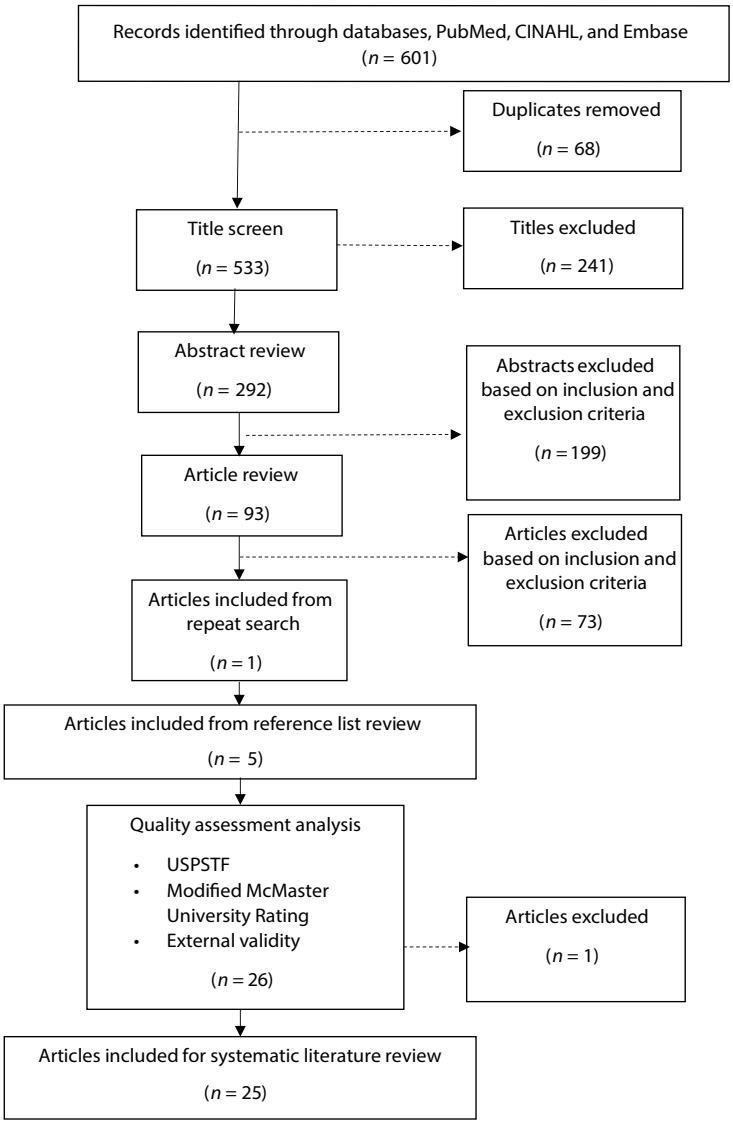

Figure 1. Flow diagram illustrating search strategy and scoping review stages

a population-based screening program. Each study was graded on the following parameters: good, fair, and poor. This rating was based on a set of conditions which considered the plausibility of the study, similarities in study population, test conditions, and social and/or environmental factors [49]. Studies were excluded if external validity was considered poor. Each study was reviewed independently by two separate reviewers (AC and RB) to minimise the potential for errors in judgement [53].

Data synthesis

Results were extrapolated in reference to the research questions and entered into a database. Through this process, two groups of studies were identified (Group 1 and Group 2). The first group was representative of findings within UNHS programs. The second group detailed outcomes of children who received UNHS, but where the outcomes reflected the protocol used by the study, not the protocol of the hospital's practice. For the purpose of this paper, these two groups are discussed separately to ensure that the findings and recommendations accurately reflect the data. Although the type of screening method was not included within the research questions, the details are included in the result tables (see Table 2) for reference against UNHS programs.

Results

From the initial search, 601 titles were obtained, of which 68 duplicates were removed, leaving 533 titles. Results from the title screen yielded 292 studies. Abstract screen was then conducted with reference to the inclusion and exclusion criteria, leaving 93 studies (PubMed 42, CINAHL 10, Embase 41). From the 93 full studies, 20 met the inclusion criteria. A repeat search conducted on 8 March 2018 yielded one additional study. Review of the reference lists provided a further 5 studies, resulting in 26 studies (see Figure 1). Both reviewers agreed on the studies included for

Table 1. USPSTF hierarchy of research design

I	Properly conducted randomized controlled trial (RCT)
II-1	Well-designed controlled trial without randomization
II-2	Well-designed cohort or case-control analytic study
II-3	Multiple time series with or without the intervention; dramatic results from uncontrolled experiments
III	Opinions of respected authorities, based on clinical experience; descriptive studies or case reports; reports of expert committees

analysis. One study was excluded due to inadequate ratings on the adapted McMaster score and USPSTF rating, leaving 25 studies for final review. All publications included in the review were rated as level II-2 (see Table 1).

What is the prevalence of conductive hearing loss in UNHS programs?

There were 18 studies which reported on the prevalence of CHL: 14 were directly representative of UNHS programs (Group 1) while 4 were representative of children who received UNHS but the outcomes reflected the study protocol (Group 2). All the literature was published between 1996 and 2018, which was considered appropriate in the historical context of newborn hearing screening.

Prevalence of CHL within UNHS programs (Group 1) ranged from 0.4% to 64.5% (see Table 2). Data collection periods ranged from 1 to 8 years. The number of screened children ranged from 2,018 to 1,392,427 and children seen at audiology ranged from 56 to 75,877. Age at audiology was 34.7 weeks gestational age to 13 months, with the majority seen in the first 2 months of life. Studies with the highest reported prevalence of CHL (>20%) had sample sizes of 76 to 211 seen at audiology departments. Studies with lower prevalence (<20%) of CHL had samples of 56 to 7,587, with most samples sizes above 300, indicating a trend for larger samples to yield lower prevalence of CHL. Eight of the studies reported a higher proportion of CHL in comparison to PCHL, seven of which used OAEs in their screening methods. No observable trends were evident when examining program locations or data collection time periods.

The prevalence of CHL of children in Group 2 ranged from 0.8% to 36.8% (see Table 2). Data collection periods ranged from 1 to 6 years. The number of children screened was reported in only one article (n = 260) and the number of children seen at audiology departments ranged from 38 to 5,282. Age at assessment ranged from 10 weeks to 6 months. Like Group 1, smaller samples tended to yield higher prevalence of CHL. Three out of the four studies reported a higher proportion of CHL in comparison to the proportion of PCHL, with all of these studies using automated auditory brainstem response (aABR) for screening. Bielecki et al. [54], who reported the lowest prevalence of CHL (0.8%) and the largest sample size (n = 5,282), used a two-stage TEOAE screening protocol. As only four studies were included in this group, no observable trends were identified when reviewing program locations. There was a tendency for shorter data collection periods (1–2 years) to yield higher rates of CHL in comparison with extended study periods.

Are there any sub-classifications of CHL applied within UNHS programs?

Two studies discussed sub-classifications of CHL and were reflective of UNHS protocol (Group 1). The first, a retrospective cohort study of 340 infants referred through UNHS in the Netherlands, reported CHL as 'genetic' if the child had a syndrome associated with CHL, such as craniofacial anomalies or if the loss was associated with causes other than OM [29]. The second study conducted by Jordan and Sidman [55] reported CHL as a 'permanent hearing loss' on point of referral for hearing aid fitting. However, this study relied on aABR testing for identification of hearing loss, which may over represent the incidence of hearing loss in this cohort as this is a screening, as opposed to a diagnostic, tool. This tool is also rarely used in UNHS programs for diagnostic purposes, which limits the application of these findings to broader UNHS programs.

How is CHL assessed and what onward referrals are made within UNHS programs?

Fourteen studies reported on the audiological assessment of children identified with a CHL (see Table 3). Five studies reported on diagnosis and management pathways reflective of UNHS programs (Group 1) and nine reported findings reflective of the study protocol (Group 2).

In Group 1, case history was reported in only one study [23] and otoscopy was not reported in any of the studies. Tympanometry was reported in four of the studies [24,29,55–56]. Two studies stated that 1000-Hz tympanometry [24,56] was used. Of these, one reported the use of 226-Hz tympanometry [55] and the other reported the use of 1000-Hz and/or 226-Hz tympanometry [29]. Three studies [23–24,55] related the use of behavioural audiometry (pure tone audiometry (PTA), play audiometry and visual reinforcement audiometry (VRA) in the determination of hearing loss. One of these studies reported thresholds ≥25 dB HL to be considered a hearing loss on behavioural measures [55].

TEOAEs and DPOAEs were described in all five studies. Liu & Liu [56] reported TEOAE screening with a pass criterion of ≥3dB SNR. Aithal et al. [24] reported on the use of diagnostic TEOAEs with a pass criteria of ≥6 dB SNR. Two studies reported the use of DPOAEs and provided their pass criteria. Liu & Liu [56] reported a pass criteria of ≥5 dB SNR at each frequency, while Jordan & Sidman [55] reported a pass at three of the five frequencies tested between 2 and 8 kHz, including 4 kHz.

Electrophysiological assessment was reported in four of the studies [23,24,29,56]. Of these, one article cited the use of auditory steady-state response (ASSR) in conjunction with click and tone-burst ABR [24]. The remaining three studies reported on click ABR assessment only. The pass criterion for ABR was reported in one article as ≤30 dB nHL [24]. One article in this group reported use of additional ABR analysis such as absolute wave latency, wave identification, or amplitude measures [24].

Table 2. Prevalence of hearing loss across universal newborn hearing screening programs. Blue background indicates use of UNHS protocol (Group 1); white background indicates protocol specific to study (Group 2)

Author	Protocol	Program location	Screening method	Study period	Screened population
Chen et al. [62]	Study	Taiwan (Taipei)	AABR	Jan 1993 – Jan 1995	260
Colella-Santos et al. [57]	Study	Brazil	AABR	Feb 2009 – Mar 2010	
Colella-Santos et al. [59]	Study	Brazil	AABR	Mar 2011– Apr 2013	
Bielecki et al. [54]	Study	Poland	2-stage TEOAE	2003–2009	
Friderichs et al. [72]	UNHS	South Africa	2-stage DPOAE	Aug 2008 – Mar 2010	2,018
Cox and Toro [73]	UNHS	US (MA)	DPOAE then AABR	Apr 1996 – Dec 2000	7,415
O'Connor et al. [3]	UNHS	Ireland	TEOAE and AABR	Apr 2011 – Apr 2012	11,738
Liu & Liu [56]	UNHS	China	TEOAE only	Oct 2006 – May 2008	11,894
Bevilacqua et al. [23]	UNHS	Brazil	2-stage TEOAE	3 years (dates not stated)	12,667
Wroblewska-Seniuk et al. [25]	UNHS	Poland	OAE	Jan 2010 – Dec 2013	27,935
Mehl & Thomson [7]	UNHS	US (CO)	TEOAE and AABR	1992–1996	41,796
Mehl & Thomson [63]	UNHS	US (CO)	OAE then AABR	1999	63,590
Spivak et al. [4]	UNHS	US (LI)	OAE then AABR	2001–2006	114,121
Szyfter et al. [74]	UNHS	Poland	not reported	2003–2006	1,392,427
Aithal et al. [24]	UNHS	Australia, N Qld	AABR	Aug 2004 – Mar 2009	
Pereira et al. [6]	UNHS	Brazil	TEOAE and CPR	2000–2002	
Holster et al. [29]	UNHS	Netherlands	OAE then AABR	Sep 1999 – Oct 2007	
Boone et al. [27]	UNHS	US (AR)	TEOAE with medical	Aug 1999 – Oct 2001	

Key: AABR (automated auditory brainstem response); OAE (otoacoustic emissions); DPOAE (distortion product otoacoustic emissions); TEOAE (transient evoked otoacoustic emissions); HWNL (hearing within normal limits); SNHL (sensorineural hearing loss); CHL (conductive hearing loss); ANSD (auditory neuropathy spectrum disorder); ND (not determined)

Table 3. Audiological tests conducted for CHL through UNHS

Article	Program location	Protocol	Otoscopy	ASSR	ABR	ABR-click	ABR-TB	ABR pass mark (re nHL)	ABR absolute latency
Karzon & Cho Lieu [58]	US (MO)	Study			X	X	X	≤ 20 dB	
Colella-Santos et al. [57]	Brazil	Study			X	X		≤ 30 dB	X
Chen et al. [62]	Taipei	Study			X	X		≤ 35 dB	
Szabo et al. [67]	US (CT)	Study							
Pereira et al. [13]	Brazil	Study							
Bielecki et al. [54]	Poland	Study	X		X	X	X		X
Doyle et al. [61]	US (CA)	Study							
Doyle et al. [60]	US (CA)	Study			X				
Colella-Santos et al. [59]	Brazil	Study	X		X		X	≤ 30 dB	X
Jordan & Sidman [55]	US (MN)	UNHS							
Holster et al. [29]	Netherlands	UNHS			X	X			
Bevilacqua et al. [23]	Brazil	UNHS			X				
Aithal et al. [24]	Australia	UNHS		X	X	X	X	≤ 30 dB	
Liu & Liu [56]	China	UNHS			X	X			

Key: ABR (auditory brainstem response); ABR-TB (ABR tone-burst); DPOAE (distortion product otoacoustic emissions); TEOAE (transient evoked otoacoustic emissions); Tymp (tympanometry); VRA (visual reinforcement audiometry); Play (play audiometry); PTA (pure tone audiometry)

No. seen at audiology	Age at audiology	No. with hearing loss	HWNL (%)	PCHL (%)	CHL (%)	Mixed	ANSD	Other/ ND
38	3–4 months			8 (21.1%)	14 (36.8%)			
38	1–6 months		16 (41.1%)	10 (26.3%)	12 (31.6%)			
929		51	29 (3.1%)	7 (0.8%)	14 (1.5%)		1 (0.11%)	
5,282	10 weeks (median)		5002 (94.7%)	240 (4.5%)	40 (0.8%)			
56	13.5 weeks (mean)			3 (5.4%)	6 (10.7%)			
138				23 (16.7%)	28 (20.3%)			
525	10 weeks (median)	15		8 (1.5%)	2 (0.4%)	3 (0.6%)	2 (0.4%)	
109	3 months	68		16 (14.7%)	31 (28.4%)	21 (19.3%)		
366	1 month – 1 year & 1 month		312 (85.3%)	11 (3.0%)	43 (11.8%)			
N/A		109		38 (34.9%)	56 (51.4%)	15 (13.8%)		
1,296				94 (7.3%)	32 (2.5%)			
1,283	2.1 months (median)	86		76 (5.9%)	21 (1.6%)			
1,222	8.7 weeks (median)		855 (70.0%)	211 (17.3%)	129 (10.6%)			
75,877		2,485		1574 (2.1%)	911 (1.2%)			
211			117 (55.5%)	26 (12.3%)	47 (22.3%)	4 (1.9%)		17 (8.1%)
1696			77%	6%	13%		4%	
340	34.7 (well-baby nursery) –39.6 (NICU) median		72 (21.2%)	197 (57.9%)	69 (20.3%)			2 (0.6%)
76	2 weeks – 10 months			23 (30.3%)	49 (64.5%)			

ABR wave identification	ABR wave amplitude	ABR interpeak latencies	TEOAE	DPOAE	Tymp 1000 Hz	Tymp 226 Hz	Reflex	VRA	Play	PTA	Cochleo-palpebral reflex
				X	X						
X	X	X	X		X						
			X			X		X	X	X	X
X		X		X	X		X				
			X		X	X		X			
X	X	X	X		X		X				
				X		X		X	X	X	
			X	X	X	X					
			X					X	X	X	-
			X		X			X	X	X	
				X	X						

Two of the studies in this group monitored the hearing and middle ear status of children identified with a CHL. Bevilacqua et al. [23] reported follow-up at 204 days (7 months) and 895 days (29 months) post-audiology assessment, while Aithal and colleagues [24] provided a follow-up diagnostic assessment at 6–8 weeks post initial audiology assessment. Five studies also reported onward referrals in the management of CHL. Doyle et al. [61] included review by a medical physician [61], while four studies reported on referral for specialist management [24, 54, 57, 58].

Most studies with evidence relating to the audiological assessment and onward referrals for CHL were reflective of the study protocol (Group 2). This may have been due to the study purpose, which was often evaluating the efficacy or hearing outcomes of UNHS programs. Unlike the first group, two studies reported on the use of otoscopy [54,59]. Four studies reported on the use of 1000-Hz tympanometry [13,57–59], and one article reported on the use of 226-Hz tympanometry. The article by Pereira et al. [13] was the only study to use behavioural audiometry (VRA, play, or PTA) and the cochleopalpebral reflex. One article by Bielecki et al. [54] used reflex testing ranging from 500 to 4000 Hz.

Three of the studies used TEOAEs [13,57,59], while two studies [54,58] used DPOAEs. Doyle and colleagues [60] used TEOAE screening with a pass criterion of ≥3 dB SNR. One study reported on the use of diagnostic TEOAEs with a pass criterion of ≥3 dB [61].

Six studies reported the use of electrophysiological assessments [24,54,57–60]. Five studies [54,57–59,62] reported using click ABR, and three studies [54,58,59] used tone-burst ABR in the determination of hearing thresholds. In Group 2, greater detail was provided on the ABR pass mark in comparison to the first group, with this ranging from ≤20 dBnHL to ≤35 dBnHL. Further analysis by electrophysiological assessments was also evident, with four studies [54,57,59,61] using wave latency, wave identification, and amplitude measures in the determination of hearing thresholds. Two studies defined the requirement for an air-bone gap (>10 dB) in the determination of CHL [58,62]. No studies in this group reported follow-up or management pathways following the identification of CHL.

What is the current medical management of children identified with a conductive hearing loss within UNHS programs?

Five studies in this review examined current medical or specialist management of children identified as CHL from UNHS (Group 1). Boone et al. [27] reported that a 'watch and wait' approach was often adopted, followed by the prescription of oral antibiotics for prolonged presentation of OM (>12 weeks). Insertion of tympanostomy tubes was only considered in cases of persistent OM and to aid accurate audiological diagnosis. However, diagnosis was conducted through an alternative protocol of TEOAEs and specialist evaluation. Mehl and Thomson [63], who reviewed the UNHS program in Colorado, USA, recommended ventilation tubes for children with CHL associated with congenital factors (craniofacial anomalies or syndrome). Aithal and colleagues [24], who examined outcomes of infants referred through UNHS in Queensland, Australia, reported medical management consisting of watchful waiting, oral antibiotics, myringotomy, or tympanostomy tube insertion. Two studies reported an additional medical screening stage post newborn hearing screening and prior to referral for diagnostic audiology assessment. This additional stage involved otolaryngologic examination to determine the condition of the external auditory canal and tympanic membrane [6,23,54].

Discussion

Given the high prevalence of CHL within paediatric populations, many children are identified with a CHL through UNHS programs. Given that to date this has not been the target condition for UNHS programs, there is limited evidence relating to the audiological diagnostic assessment and management of these children. The present study therefore aimed to review detection of CHL within a UNHS context, including determining the prevalence and classification of CHL, as well as audiology and medical management.

What is the prevalence of CHL within UNHS programs?

Highly variable prevalence rates for CHL were evident across the 18 studies, with rates ranging from 0.4% to 64.5%. Upon group comparison, Group 1, which was representative of UNHS program protocols, had a greater range of CHL prevalence (0.4–64.5%) than studies in Group 2 (0.7–36.8%). While only four studies were included in Group 2, the tendency for this group to have more consistency in prevalence rates may be explained by the study protocol, as the majority were validating screening protocols for the identification of CHL. This could indicate greater rigour in the study design and selection of audiology assessments to identify CHL. Overall, investigation of the sample size of children seen at audiology produced an observable trend, with larger samples yielding lower prevalence of CHL.

There was no observable trend between reported age of diagnosis and prevalence rates observed in either group of studies. Most children were seen in the first 3 months of life. However, comparisons were limited, as age was often reported as a range (e.g., 1–6 months). Without details of age of diagnosis, investigation into the peak prevalence of CHL in the first year of life cannot be addressed. The prevalence of OM in the first year of life is significant (up to 73%), with nearly all children affected by 3 years of age [13,14]. Further research linking peak prevalence of OM to CHL could contribute to a better understanding of the impacts of OM on hearing in the first few years of life and the best opportunities for identification and intervention.

Eight of the studies in Group 1 reported higher prevalence of CHL in comparison to PCHL. This may indicate a genuine difference in prevalence rates, or may be related to the choice of screening technology, as seven of eight studies reported OAEs as the screening method. Indeed,

the general literature suggests that OAE screening yields a higher referral rate or more children referred without the target condition (PCHL) [64]. However, a thorough investigation into the effect of an OAE screening protocol on rates of children referred with CHL within UNHS has not been adequately addressed. While there has been support for the use of diagnostic TEOAEs in the identification of CHL in general paediatric populations [65], there has been some disagreement over the ability of TEOAEs to effectively detect middle ear dysfunction [66]. Further examination into the type of hearing loss identified (CHL vs PCHL) by screening protocol could contribute greatly to this body of research.

Explicit patient characteristics were investigated in only three studies (Group 2), which examined the hearing outcomes of infants who had a previous admission to NICU. All studies reported higher prevalence of conductive hearing loss in comparison to PCHL. Further investigation into pre- and post-birth factors associated with admission to NICU could be highly beneficial within a UNHS program. While evidence for the use of high risk indicators for PCHL, such as cleft palate and syndromes, has been demonstrated in the literature [25,55,67], a risk factor registry specific to CHL has yet to be published for application within UNHS programs.

Are there any sub-classifications of CHL that guide specific interventions within UNHS?

Overall, two sub-classifications of CHL were evident in the literature and were reported in Group 1 studies. One study reported CHL as "permanent" if hearing aids were prescribed [55], while the other introduced the term "genetic CHL" if the hearing loss was attributed to congenital factors, such as syndromes or craniofacial anomalies [29]. These minimal findings suggest that the classification of CHL may be influenced by several patient characteristics relating to the cause, severity, and longevity of CHL. Given the number of aetiologies resulting in chronic middle ear dysfunction and resulting CHL [21,22], development of sub-classifications for CHL may be beneficial within UNHS. An absence of these sub-classifications places children likely to develop chronic CHL at risk of further developmental delays due to delayed or inappropriate interventions.

How is CHL assessed and what onward referrals are made within UNHS programs?

Evidence for the audiological assessment and ongoing management of CHL within UNHS was reported in 14 studies. Overall, there was no standard test battery to assess CHL, with testing often following protocols for the detection of PCHL. The tests included: case history, TEOAEs, DPOAEs, tympanometry (1000 Hz and 226 Hz), acoustic reflexes, cochleopalpabral reflex, tone-burst and click ABR (air conduction and bone conduction), ASSR, and behavioural audiometry (PTA, Play, and VRA).

Otoscopy was only reported in two studies, both of which were reflective of the study protocol. Similarly, acoustic reflexes were only reported in two studies and were also included in the study protocol group. The cochleopalpabral reflex was used in two studies, represented in both Groups 1 and 2 [13,23]. An additional search of the literature yielded very few recent studies on the cochleopalpabral reflex. Both studies discussed the application of this test in Brazilian paediatric populations [68,69] and cannot be translated into other UNHS screening program protocols.

Otoacoustic emissions were used in 8 of the 14 studies. Three studies in Group 1 used TEOAEs, two used DPOAEs, and one study used both. Two studies in Group 2 used DPOAEs and three used TEOAEs. When combining both groups, studies that used TEOAEs reported prevalence of CHL from 1.5–31.6% ($n = 4$). Three of these studies also reported higher prevalence of CHL over PCHL. Where DPOAEs were used and prevalence was reported, CHL rates were 0.8% and 28.4%. Overall, there was no observable trend between TEOAE and DPOAEs to identify more or less CHL. This is not consistent with the general literature where an inconsistency in the efficacy of TEAOEs to identify CHL has been established [65,66], while the application of DPOAEs as a predictor of even mild CHL has been documented [70]. Identification of the best OAE method to identify CHL could introduce significant efficiencies into UNHS programs worldwide.

ASSR was the least commonly used electrophysiolocal assessment for both groups, with only one study in each group using this assessment method. An absence of this method may be explained due to its poor agreement with mild to moderate behavioural thresholds [71]. Auditory Brainstem Response (click and tone-burst) was used in most of the studies. However, due to the limited information provided and high variability between study findings, it was not possible to investigate any trends between pass criteria and prevalence rates.

Audiological monitoring for CHL was only reported in Group 2, ranging from 2 to 29 months post diagnostic assessment. Onward referrals included GPs and otolaryngologists. Unfortunately, no information was reported on the number of assessments or age of identification, an omission which means that the resources allocated to manage this cohort or the natural progression of the disease cannot be quantified. This review also revealed an absence of referrals for hearing amplification, counselling support services, or additional developmental support, suggesting an absence of protocols in this area. As research indicates that children identified with CHL are less likely to be fitted with hearing amplification within an acceptable time-frame and are more likely to disengage with supportive services [4], the current review highlights the importance of further investigation into the progression of CHL and the development of referral guidelines for non-medical management options.

What is the current medical management of children identified with a conductive hearing loss within UNHS programs?

This question produced the smallest body of evidence. All studies were in Group 1 and were representative of UNHS protocol. Specialists' management comprised an observation period, prescription of oral antibiotics, and insertion of tympanostomy tubes [27]. The insertion of tympanostomy

tubes was often recommended after an extended duration (>12 weeks) of OM, where an accurate audiological diagnosis was yet to be obtained, or if congenital factors associated with CHL were identified (craniofacial or syndromes) [27]. Surprisingly, other common management options for OM detected through UNHS were not found in this review, including the prescription of topical antibiotics, steroids, surgical procedures such as myringotomy, or referral for hearing aids. Further research into the current specialist management of CHL, including outcomes and options for care pathways, is needed in order to better understand the medical management of CHL within UNHS programs.

Limitations

Several limitations were evident in this literature review. Overall, a small number of studies met the inclusion and exclusion criteria, which limited the comparison of these results to large-scale UNHS populations. Through the review, two distinct groups of studies were identified: those representative of UNHS program protocol (Group 1), and those representative of study protocol (Group 2). This reduced the findings directly relevant to existing UNHS protocols to only 15 studies. Furthermore, a notable difference between population characteristics, sample sizes, and collection periods were evident among the studies. Most of the evidence obtained in this review was derived from inferential findings, often reported only in the research methodology. Finally, a significantly low yield of studies examining specialist management of CHL within a UNHS context was evident. Therefore, caution must be practised when applying these findings to screened populations.

Conclusions

The results from this systematic literature review demonstrate a significant gap in the literature with regards to identification and management of CHL within UNHS programs. The review identified the following issues. (1) The prevalence of CHL within UNHS is highly variable, ranging from 0.4% to 64.5%; overall, results suggest that higher samples yield lower prevalence of CHL. (2) Two sub-classifications of CHL were infrequently reported within UNHS programs. (3) The audiological management of CHL within UNHS involved many and varied audiological assessments, typical in the assessment of PCHL. Limited evidence was obtained as to the most appropriate test battery for the identification of CHL within UNHS. Limited evidence of ongoing audiological management or onward referrals was found. (4) Very little evidence was found on the specialist management of CHL within UNHS programs. Management options included the prescription of oral antibiotics, a watch and wait approach, or surgical interventions such as tympanostomy tubes. No alternative medical pathways for children identified from UNHS were evident.

In general, the impact of CHL and the understanding of appropriate assessment and early interventions for children with CHL within UNHS programs is unknown. Further investigation to address these research questions is recommended to: (1) clarify the true prevalence of CHL, including a method to identify children at risk of chronic CHL; (2) establish sub-classifications of CHL within UNHS programs to reflect the cause, predicted longevity, and risk of developmental delay for children with CHL; (3) investigate the audiological management of CHL within UNHS, including appointment numbers, tests conducted, review time-frames, and associated outcomes; (4) analyse specialists' management of CHL within UNHS, including types of assessments or surgical procedures and outcomes; and (5) develop a 'best practice' model which identifies the appropriate care pathways for children identified with CHL within UNHS programs.

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