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REVIEW ARTICLE Open Access

Cervical and ocular vestibular evoked myogenic potential in children with sensorineural hearing loss with and without cochlear implant: a systematic review

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Abstract

Background: Children with sensorineural hearing loss (SNHL) form a major chunk of the population with hearing difficulty. Since the auditory system is close to the vestibular system and shares the common fluid, the abnormality in the auditory system might impair the functioning of the vestibular system.

Main body of the abstract: The present systematic review aimed at studying the application of cervical and ocular vestibular evoked myogenic potential responses (cVEMP and oVEMP) during assessment and rehabilitation of children with SNHL with and without a cochlear implant. A systematic search was done across databases on cVEMP and oVEMP findings in children with SNHL. Out of 92 articles retrieved, 21 articles were found to be appropriate as per our inclusion criteria. Significant vestibular abnormality was seen in children with SNHL as shown on cVEMP and oVEMP test reports. The cVEMP and oVEMP abnormalities seen were more in children with SNHL using a cochlear implant. Pre- and post-implantation studies showed a significant reduction in measures of cVEMP and oVEMP after implantation.

Short conclusions: Studies in the last decade reported abnormal cVEMP and oVEMP response in children with SNHL with and without cochlear implantation. cVEMP and oVEMP response is also associated with poor motor development in children with SNHL. Thus, emphasis should be given to assessing vestibular functioning in children with SNHL to rehabilitate them early in life.

Keywords: Vestibular-evoked myogenic potential, Cochlear implant, Sensorineural hearing loss, Vestibular system

Background

Sensorineural hearing loss (SNHL) is commonly seen as hearing loss due to the inner ear or the vestibulocochlear nerve damage. Literature review suggests SNHL accounts for 90% of reported hearing loss which is permanent and varies in severity from mild to profound hearing loss [1].

Recent studies reported the importance of studying the relationship between peripheral vestibular function and cochlear function, underlined by the fact that the cochlea and vestibular systems are anatomically, phylogenetically, and functionally related [2]. Hence, disturbances in cochlear function, which can result in SNHL, could accompany vestibular impairment because of sharing the membranous labyrinth of the inner ear with the vestibule. Prenatal, natal, or postnatal injury may cause damage to one or both the systems leading to SNHL [3–5].

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Early detection of vestibular dysfunction in the pediatric population is important. If the vestibular dysfunction goes undiagnosed and untreated, this would result in significant motor impairment [6] and inability to maintain stable gaze [7], thus affecting their scholastic performance. Although a small proportion of these individuals with SNHL exhibit concurrent vestibular involvement, it is important to identify the impairment and rehabilitate them [8]. From birth onwards, auditory stimulation directs and intensifies visual orientation behavior in infants and the infant's earliest responses to auditory stimuli include the visual-motor behavior of moving the eyes or head to localize sound. Therefore, it has been suggested that lack of early auditory input could contribute to motor delays in children with hearing impairment [9]. Several published reports also suggest the influence of vestibular dysfunction on the gross motor development of individuals with SNHL [10, 11]. One of the audiological tests to assess the functioning of the vestibular system is vestibular evoked myogenic potential (VEMP).

VEMP is a short latency electromyographic and neurophysiological assessment technique that can be recorded from various muscles in response to the acoustic stimulus [12, 13]. There are two types of VEMP, cervical VEMP (cVEMP) and ocular VEMP (oVEMP). oVEMP measures the functioning of the utricle and superior vestibular nerve whereas cVEMP measures the functioning of the saccule and the inferior vestibular nerve [14]. oVEMP is an excitatory myogenic response recorded mainly from the contralateral extraocular muscles [15]. cVEMP is an ipsilateral inhibitory response recorded from the sternocleidomastoid muscle [15]. VEMP recording is considered a simple and rapid method which is patient-friendly and easily implementable in a clinical setup [16]. Both cVEMP and oVEMP are found to be reliable tools in children [17–19].

The key to the management of children with SNHL is early diagnosis and early intervention which primarily includes amplification devices and cochlear implantation along with speech and language intervention to facilitate the child's overall communication ability. Children with SNHL show vestibular system dysfunction as highlighted by the published reports. Many of the researchers have shown abnormal cVEMP and oVEMP results in children with SNHL [8, 20–23]. Reports also suggest vestibular dysfunction in children with SNHL using cochlear implants [24–26]. Since there are many published reports in the last decade highlighting the cVEMP and oVEMP results in children with SNHL, we aimed to compile all the reports relevant to children with SNHL with and without CI and VEMP results. This systematic review aims to explore the application of VEMPs during the assessment and rehabilitation of children with SNHL with and without CI.

Methods

Study design

We performed a systematic literature review of articles published on cVEMP and oVEMP in children with SNHL with and without cochlear implants (CI). A written study protocol was prepared following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement [27].

Search strategy

We systematically performed a search in PubMed, Google Scholar, Wiley Online Library, and Google databases for the cVEMP and oVEMP data in the last 10 years (January 2010 to December 2020). The collective search strategy was incorporated and executed independently by both the researchers with no imposed data limits upon the search. In December 2020, a structured computerized search was performed. The keywords, phrases, and MeSH terms searched included the following: "cVEMP in Children with SNHL," "oVEMP in Children with SNHL," "cVEMP in Children using Cochlear implants," and "oVEMP in Children using Cochlear implants." The search was limited to articles published in English. Furthermore, articles with systematic review and meta-analyses on this topic helped us to identify additional articles, which got missed in our database search. Table 1 shows the number of articles retrieved on cVEMP and oVEMP in children with SNHL with and without CI.

Inclusion and exclusion criteria

The inclusion criteria included a sufficient and accurate description of pathologies, their clinical features, and VEMP recording. Studies performed on children up to the age of 18 years were only considered. The articles on cVEMP and oVEMP in children with SNHL with and without CI were included. The exclusion criteria included articles published before the year 2010, repeated studies or overlapping citations in databases, studies other than in children with SNHL, and test findings other

Table 1 Number of articles retrieved on cVEMP and oVEMP in children with SNHL with and without a cochlear implant

Database	cVEMP articles	oVEMP articles	Total
PubMed	13	14	27
Google Scholar	08	07	15
Wiley Online Library	03	03	06
Google databases	15	10	25

than VEMP. All the articles considered had investigated cVEMP/oVEMP/both in children with and without CI.

Data extraction

The literature search was independently conducted by both the reviewers. The articles collected by both the reviewers were analyzed for study design, participant's characteristics, test measures used, and the result of the tests. The inclusion and the exclusion criteria were kept in mind before considering the articles for review, and in case of discrepancy in opinion between the reviewers, the full-text article was reviewed again by both the reviewers and a mutual decision was taken. Figure 1 shows the *PRISMA* flow chart of the literature review process considered in this study.

Ethical consideration

The study was approved by the Institutional Ethical Committee. Informed consent from the participants was not applicable as no human subjects were directly involved for this study as this is a review article.

Results

Overall, the primary search yields a total of 92 articles. Independent screening of abstracts and titles of the articles was performed. The inclusion and exclusion principles adopted in the study resulted in 21 articles for cVEMP and oVEMP. Some of the articles investigated both the cVEMP and oVEMP responses while others had either of the cVEMP or oVEMP responses.

cVEMP and oVEMP in children with SNHL

Many of the researchers have studied the characteristics of cVEMP and oVEMP in children with hearing loss [8, 20, 22, 28–30]. Table 2 shows the summary of the cVEMP and oVEMP studies on children with SNHL. Singh et al. recorded cVEMP response in 15 children with severe to profound hearing loss and 10 children with normal hearing [22]. Of the 15 children with SNHL, cVEMP response was absent in 2 (13%) of the individuals. The latencies of P13 and N23 responses did not show a significant difference whereas peak-to-peak amplitude showed a significant difference across the groups. Said showed absent cVEMP response bilaterally in more than 45% of children with SNHL, 12% in unilateral SNHL, and delayed cVEMP

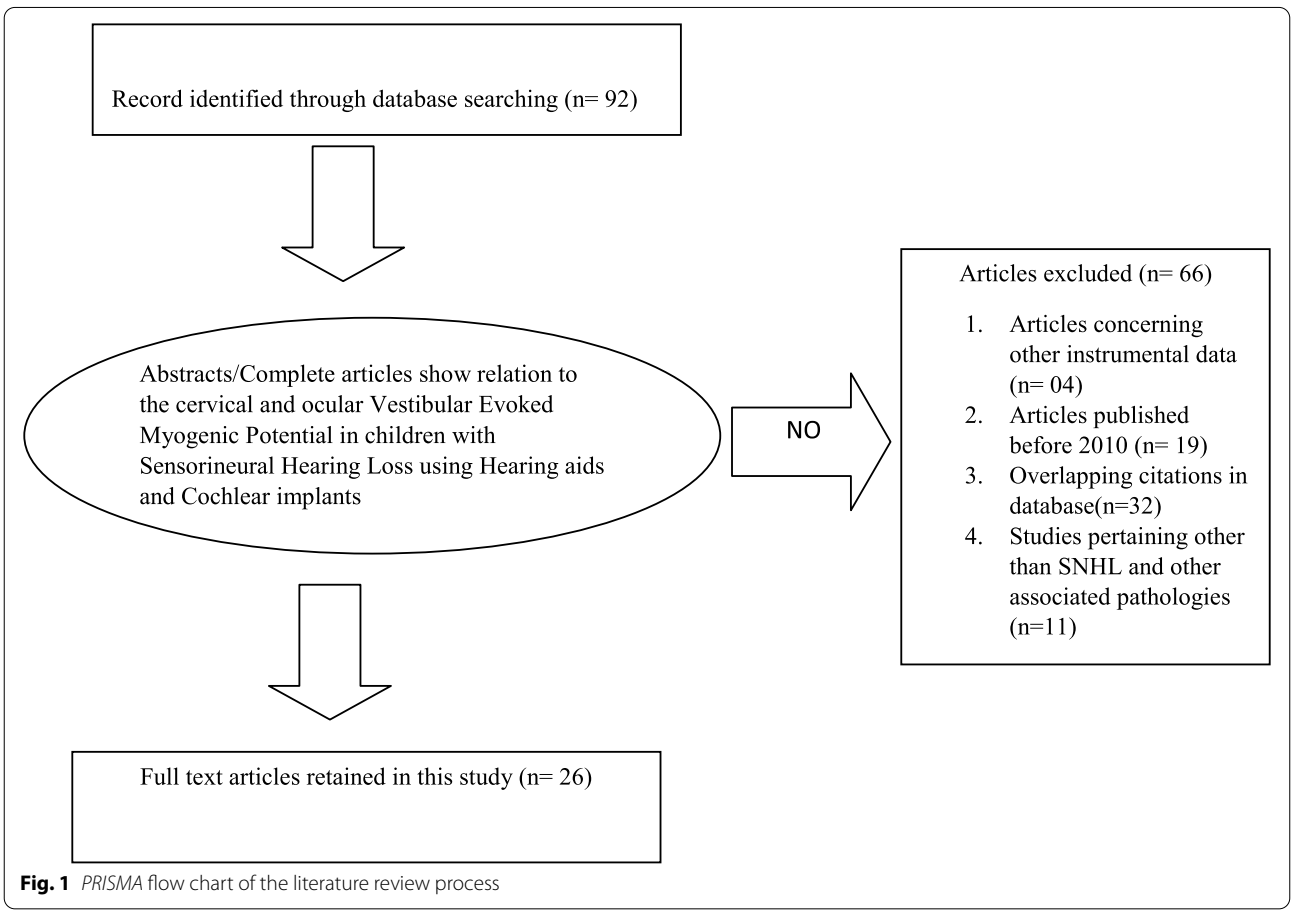

Fig. 1 *PRISMA* flow chart of the literature review process

Table 2 Summary of cVEMP and oVEMP test results seen in individuals with SNHL

Sl. no.	Authors	Year	Tests administered	Number of participants	Significance
1	Jafari and Malayeri [33]	2011	cVEMP	30	++
2	Singh et al. [22]	2012	cVEMP	15	++
3	Kegel et al. [10]	2012	cVEMP	48	++
4	Selim et al. [31]	2012	cVEMP	55	++
5	Inoue et al. [11]	2013	cVEMP	89	++
6	Bansal et al. [21]	2013	cVEMP and oVEMP	23	++
7	Said [8]	2014	cVEMP	50	++
8	Zhou et al. [28]	2014	cVEMP	275	++
9	Xu et al. [29]	2015	cVEMP and oVEMP	43	++
10	Emami and Farahani [30]	2015	cVEMP	100	++
11	El-Badry et al. [32]	2019	cVEMP	67	++
12	Apeksha et al. [20]	2020	cVEMP and oVEMP	15	++

++ significant difference in any of the cVEMP/oVEMP test parameters between the children with normal hearing and with SNHL

responses in 20% of individuals with SNHL [8]. The percentage of abnormal/absent cVEMP response was higher in children with a higher degree of SNHL compared to a lesser degree of hearing loss [8, 31]. cVEMP abnormality seen in children with profound SNHL varied from 56% in bilateral pathology to 16% for unilateral pathology. cVEMP responses are present for the lesser degree of hearing loss and are usually absent for the profound degree of SNHL. Zhou et al. in a retrospective study found 54% of the children with different degrees of SNHL to have abnormal cVEMP responses [28].

Xu et al. reported a response rate of 58.1% and 61.9% for oVEMP and cVEMP respectively in children with profound SNHL. There was a significant reduction in amplitude and elevation in the threshold of oVEMP and cVEMP in children with profound hearing loss compared to healthy children [29]. Researchers have also observed a significant difference in cVEMP response in children with severe-to-profound hearing loss with and without auditory neuropathy spectrum disorder [30, 32]. The study suggests a positive correlation between saccular dysfunction and ANSD in children with severe-to-profound hearing. Apeksha et al. reported the absence of cVEMP and oVEMP response in 27% of the children with severe-to-profound hearing loss [20]. Even though none of the children or their parents report difficulty in performing their daily living activity, the Fukuda stepping test and the Tandem gait test showed abnormal results suggesting a compromised balance system. Kegel et al. and Inoue et al. suggested a combination of rotatory chair testing and VEMP testing to be used for children with SNHL to predict the balance performance in them [10, 11].

Based on the above-mentioned studies, it is clear that a significant number of children with SNHL show vestibular system abnormality as shown in VEMP results. The VEMP abnormalities seen were reduction in amplitude of the peaks, prolongation in latencies of the peaks, and increase in the VEMP thresholds and the majority of the children showed complete absence of VEMP response [20, 21, 33]. Researchers believe that it is the anatomic compartmentalization of vestibular end organs and the cochlea that are likely to show an abnormality in both cochlear and vestibular function. It is because of the proximity of the structures that the saccule show more significant abnormality followed by utricle and the semicircular canals in individuals with SNHL [34, 35]. Some researchers also suggested more debilitating symptoms in individuals having both cochlear as well as vestibular damage compared to individuals with only cochlear damage [36]. Children who showed abnormal VEMP response also showed significantly delayed acquisition of head control and sitting and independent walking [10, 11]. Recent studies reported abnormal VEMP findings in most of the hearing-impaired children with no reported vestibular symptoms which may be due to the inability to report or describe the dizziness as they are too young to describe it [20]. Saccular impairment in isolation might not cause vestibular disturbances in children as the input from the impaired vestibular system might get compensated by the central component. Studies also suggest that the abnormality seen on cVEMP and oVEMP responses correlates well with the degree of hearing loss [8, 31].

cVEMP and oVEMP in children with SNHL using the cochlear implant

Several researchers have explored the functioning of the vestibular system in children with SNHL using CI using cVEMP and oVEMP responses. The majority of the

published reports suggest an adverse effect of CI on the functioning of the vestibular system post-implant [23–26, 37]. Table 3 shows the summary of the cVEMP and oVEMP studies on children with SNHL using CI.

Xu et al. found the response rate of cVEMP and oVEMP to be 67.7% and 71% before the CI surgery in children with SNHL [23]. After 1 month of CI surgery, the response rate decreased to 32% and 12.9% respectively for cVEMP and oVEMP. For children in whom the cVEMP and oVEMP responses were recorded, the response showed a reduction in amplitude of peaks and elevation in thresholds suggesting significant damage to the vestibular system. In another study, VEMP response was abnormal in 60% of the children pre-operative and became 100% abnormal 6 months post-operative in the implanted ear [38]. In a group of children implanted unilaterally and planned to undergo contralateral implantation, Devroede et al. showed the presence of cVEMP response in 79% of the children pre-operatively and 62% post-operatively [39]. Thierry et al. reported normal VEMP response in six children, partial dysfunction bilaterally in four children, and asymmetrical vestibular function in two children pre-implantation. Post-implantation, six children had the same vestibular result; four children showed improvement in vestibular symptoms after surgery, and two children showed worsening of symptoms [26].

Imai et al. investigated the functioning of otolith organs before and after CI. cVEMP results showed an increase in asymmetry ratio in 5 out of 9 children 1 month postoperatively indicating deterioration in sacculo-collic response post-CI [25]. oVEMP results showed an increased asymmetry ratio for 10 out of 11 children with CI, indicating deterioration of utriculo-ocular response postoperatively. The majority of the researchers reported significant deterioration of cVEMP and/or oVEMP response in children with CI, except one of the studies [40]. Ajalloueyan et al. recorded cVEMP response in 27 children with severe to profound hearing loss [40]. The researchers reported no statistically significant difference in cVEMP response pre-post implantation.

Li et al. also observed otolith dysfunction in 35 children with CI, suggesting VEMP be one of the reliable tools to assess the functioning of otolith organs post-implantation [24]. Merchant et al. compared cVEMP and oVEMP elicited by air conduction and bone conduction stimuli to check the hypothesis that it is not the actual vestibular loss but the mechanical changes by the CI that leads to the absence of VEMP response in children with CI [37]. Bone-conduction VEMP response rate was higher than the air-conduction VEMP responses suggesting that it is the mechanical property of the peripheral system which gets altered with CI surgery and leads to an air-bone gap rather than the actual vestibular loss in children with CI [37]. Many researchers reported alteration in the intracochlear pressure and peripheral mechanics as the cause of the persistent air-bone gap in individuals with CI [41–43].

Researchers in the field have no clear justification for the effect of CI on the non-implanted side. Katsiari et al. reported either absent or abnormal VEMP responses after CI surgery on the non-implanted side of the children with SNHL [44]. Similarly, Psillas et al. also reported abnormal VEMP responses in the non-implanted ear of children with CI [38]. This finding may be supported by the hypothesis that the abnormal responses on the non-implanted side might be a consequence of abnormal input of the vestibular system on both sides. With the insertion of the CI electrodes in one ear, the vestibular input to the brain gets altered in the same ear and indirectly modifies the contralateral ear vestibular response. Despite abnormal VEMP findings seen in children with CI, none of the children reports dizziness or vertigo [20, 38]. This could be attributed to the capacity of the

Table 3 Summary of cVEMP and oVEMP test results seen in individuals with SNHL using a cochlear implant

Sl. no.	Authors	Year	Tests administered	Number of participants	Significance
1	Katsiari et al. [44]	2013	cVEMP	20	++
2	Xu et al. [23]	2014	cVEMP and oVEMP	31	++
3	Psillas et al. [38]	2014	cVEMP	10	++
4	Devroede et al. [39]	2016	cVEMP	26	++
5	Thierry et al. [26]	2015	cVEMP	43	++
6	Ajalloueyan et al. [40]	2017	cVEMP	27	--
7	Imai et al. [25]	2019	cVEMP and oVEMP	21	++
8	Li and Gong [24]	2020	cVEMP and oVEMP	35	++
9	Merchant et al. [37]	2020	cVEMP and oVEMP	27	++

++ significant difference in any of the cVEMP/oVEMP test parameters between the children with normal hearing and with SNHL using a cochlear implant

-- non-significant difference between cVEMP and oVEMP test parameters between the children with normal hearing and with SNHL using a cochlear implant

children to compensate for the peripheral vestibular loss by the process of central compensation.

The vestibular system is found to be at risk for damage following CI surgery because of the anatomical proximity of vestibular and cochlear systems [37]. The significant damage caused to the inner ear structures includes fibrosis in the vestibule, the collapse of the saccule, decrease in ganglion cells, inner ear hydrops, and scar tissues [34, 45]. Obeidat et al. reported otolith organs to be more affected than the semicircular canals [35]. Of the two otolith organs, the saccule is the most vulnerable site for damage following CI surgery than the utricle [34]. This is explained based on the proximity of the saccule to the cochlea where electrodes are inserted and can easily damage the saccule.

Conclusions

Most of the recent studies in the last decade reported abnormal cVEMP and oVEMP responses in children with SNHL. The functioning of the vestibular system gets even more impaired following CI surgery. This suggests that CI has a more negative impact on the functioning of the vestibular structures. Vestibular dysfunction seen in children with SNHL may slow down their motor development and thus can delay the motor milestones. So we must assess the child's audio-vestibular system early in life to rehabilitate them early. It is also important that we screen all the children with SNHL for vestibular impairment irrespective of the degree and cause of SNHL.

Abbreviations

SNHL: Sensorineural hearing loss; cVEMP: Cervical vestibular evoked myogenic potentials; oVEMP: Ocular vestibular evoked myogenic potentials; CI: Cochlear implant; PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-Analyses; ANSD: Auditory neuropathy spectrum disorders.

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Authors' contributions

KA and DD performed the independent systematic title and abstract search based on the selection criteria, collected the study data independently, and analyzed these data. DD was a major contributor in writing the manuscript. KA and DD both read and approved the final manuscript.

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Availability of data and materials

The datasets used and analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

Obtained from the JSSISH ethical committee with reference No. JSSISH/2021/P8. Informed consent from the participants was not applicable as no human subjects were directly involved for this study as this is a review article.

Consent for publication

Not applicable.

Competing interests

The authors declare that they have no competing interests.

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