



Review Article

Vestibular rehabilitation exercises programs to improve the postural control, balance and gait of children with sensorineural hearing loss: A systematic review



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ABSTRACT

Background: Several studies have demonstrated that children with sensorineural hearing loss (SNHL) exhibit postural instabilities, as well as balance and gait disorders, due to the vestibular dysfunction that they are prone to display as a consequence of inner ear injury. Thus, some experiments have proposed vestibular rehabilitation exercises programs as a treatment to improve these motor skills in children with SNHL.

Objective: Assess the evidence quality of the trials that used vestibular rehabilitation exercises programs to improve the postural control, balance and gait of children with SNHL.

Methods: This is a systematic review that surveyed articles in nine databases, published up to July 4, 2019, in any language, using the following inclusion criteria: (1) Randomized or quasi-randomized controlled trials. (2) Participants of both groups with clinical diagnosis of SNHL, aged up to 12 years old, with no physical problems, cognitive or neurological impairments, except the vestibular dysfunction. (3) Using vestibular rehabilitation exercises programs to improve the following outcomes: postural control, balance and/or gait.

Results: Six experiments, including 153 children, met the inclusion criteria of this systematic review. Two randomized controlled trials (45 children) on the postural control exhibited low evidence quality and four others; three randomized and controlled trials (90 children) on the balance and one quasi-randomized (18 children) on the gait demonstrated very low evidence quality, respectively.

Conclusion: There is promising evidence that vestibular rehabilitation exercises programs improve the postural control, balance and gait of children with SNHL. However, due to the methodological limitations of the trials and low quality of current evidence on this topic, the trials results analyzed by this systematic review should be interpreted with caution. Due to the low quality of evidence observed in this review, we suggest that new trials be proposed on this topic, with better methodological quality, to prove the effectiveness of vestibular rehabilitation exercises programs to improve the postural control, balance and gait of children with SNHL.

1. Introduction

Evidence in the literature shows that children with SNHL exhibit more postural control instabilities than their normal-hearing counterparts of the same sex and age group [1–3] as well as changes in movements and postural alignment of the head [4–6] and spine [7]. Other studies reported balance disorders (static and dynamic) in this

population [8–11] and also changes in gait, such as slower speed [11], greater ground reaction force [12] and worse performance in gait-related functional tasks [13], also when compared to normal-hearing children and the same occurring in other motor skills [14–16].

These motor disorders can negatively influence the functionality, the practice of sports or recreational activities and the typical joking of the age of children with SNHL, making them not comparable to other

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Box 1
Inclusion criteria of the trials.

Design
• Randomized or quasi-randomized controlled trials.
Participants
• Children with clinical diagnosis of sensorineural hearing loss (unilateral or bilateral); • Age: ≤12 years; • No physical problems and cognitive or neurological deficits, except, vestibular dysfunction.
Intervention
• Physiotherapy interventions; • Vestibular rehabilitation exercises; • Exercises performed on soil.
Outcomes
• Primary: Postural control and balance; • Secondary: Gait.
Comparisons
• Usual or daily activities; • Leisure or recreation activities; • No intervention, or any other intervention.

children, in terms of motor performance. This could affect their social relations, causing emotional disturbances and the isolation of these children. These motor disorders in children with SNHL are due mainly to inner ear lesions, which can cause lesions in the vestibular system, since the cochlea and vestibule share the continuous membranous labyrinth of the inner ear. Thus, prenatal, perinatal or postnatal lesions can damage one or both systems [17].

The literature indicates that 20–85% of children with SNHL display peripheral vestibular dysfunctions [18–21], compromising the balance of these children. This problem may occur due to the anatomical proximity between the cochlea and vestibular apparatus, given that these two structures are peripheral sensory organs of the vestibular-auditory systems, being anatomically and functionally related in terms of innervation and vascular supply. Given the high frequency of vestibular dysfunctions, in addition to the motor and balance disorders exhibited by the children with SNHL and mentioned above, vestibular rehabilitation exercises programs are needed to treat these children [22].

Vestibular rehabilitation is a therapeutic resource applied to patients with balance disorders of vestibular origin and its proposal is based on the central mechanisms of neuroplasticity, known as adaptation, habituation and substitution, whose final objective is vestibular compensation. The exercises used in vestibular rehabilitation aim at improving vestibulovisual interaction during cephalic movements, optimizing static and dynamic postural stability under conditions that produce conflicting sensory information [23]. Vestibular rehabilitation has proven to be a safe and effective therapeutic option for treating children with peripheral vestibular disorders [24], thereby increasing interest in therapeutic approaches to minimize or reverse motor and balance disorders in children with SNHL [25].

The literature contains two systematic reviews that analyzed the effects of vestibular rehabilitation exercises only on the balance outcome of children with SNHL [26,27]. However, both reviews exhibit important methodological limitations, such as non-registration of the reviews protocols, selecting only studies published in English, did not include important databases in article searches (PEDro [26] and SCOPUS and Web of Science [27]) and both used scales to assess evidence quality of the trials, which may not adequately reflect the risk of bias of the trials. Furthermore, in one review [26], only one examiner selected the articles and assessed the evidence quality, and in the other [27] the year of publication of the articles was restricted, which is not recommended for systematic reviews.

Thus, the trials with functional outcomes, such as gait of children with SNHL, for example, have not yet had the quality of their evidence investigated, and the reviews that analyzed the evidence quality of the trials on the balance outcome presented important methodological limitations. This fact may have contributed to not reaching all the trials on this topic, justifying the conduct of this investigation. Given the above, this systematic review aimed to assess the evidence quality of randomized and quasi-randomized controlled trials that used vestibular rehabilitation exercises programs to improve the postural control, balance and gait of children with SNHL, using the best level of evidence: the GRADE system (*Grading of Recommendations, Assessment Development and Evaluation*).

2. Methods

This systematic review was conducted according to the guidelines of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) Statement [28], and its protocol was registered at PROSPERO under number (CRD42016053135) [29].

2.1. Identification and selection of the trials

The articles were identified by a search of nine databases: MEDLINE/PubMed, EMBASE, SCOPUS, LILACS, CINAHL, CENTRAL (Cochrane Central Register of Controlled Trials), Web of Science, PEDro and Google Scholar, the first search occurring on January 11, 2017. Another search was conducted on July 4, 2019, to add updated studies. There was no language or year of publication restriction of the trials and a manual search on the list of trials references was also carried out, in order to ensure that all the relevant trials on this topic was included.

The search strategies used for each database are shown in appendix 1.

The articles found in each database were analyzed independently by both examiners (Melo RS and Paiva GS), who judged their relevance after reading the titles and abstracts, in front of a computer, according to the inclusion criteria of this study (Box 1). In the first analysis, the articles were divided into eligible or excluded for this review. Studies with questionable abstracts or with potential to be included in this review were separated and read in their entirety. Disagreements regarding including or not one of the studies were resolved by the examiners and when no consensus could be reached, a third reviewer (Lemos A) was asked to arbitrate.

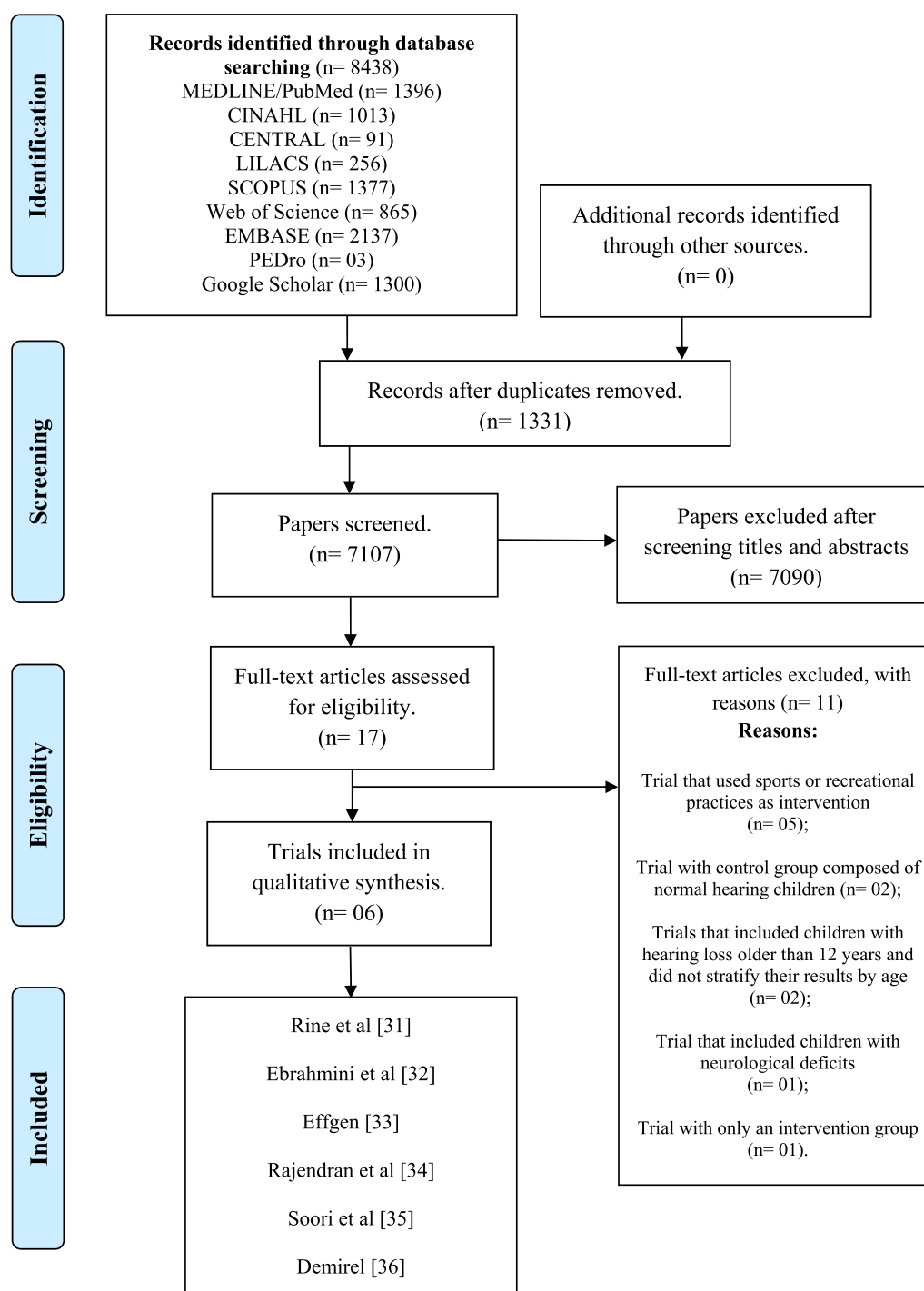


Fig. 1. Flow of studies through the review, according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA).

For cases with lack of study data, the authors of this systematic review contacted the corresponding author to obtain the necessary information. We obtained answers from all the authors of the studies that were being considered for inclusion.

2.2. Assessment of the studies characteristics

2.2.1. Quality of evidence

Quality of evidence was evaluated using the approach proposed by the GRADE system [30]. According to this proposal, five factors can interfere in evidence quality from a trial: design, risk of bias, inconsistency, indirectness and imprecision. For each of these items, evidence

was considered according to the following classification: not serious (no decrease in points), serious (decrease of 1 point) or very serious (decrease of 2 points), scored according to the risk of study bias. For the specific item 'risk of bias' of GRADE, the Cochrane instrument was used to evaluate the risk of bias of the trials, which evaluates the following items: randomization, anonymous allocation, blinding of the participants and of the outcome assessors, losses or incomplete data, selective description of outcome and others (if applicable). Each item of the risk of bias instrument was assessed as follows: low risk of bias (green), unclear risk of bias (yellow) or high risk of bias (red), according to the risk of bias presented by the trial.

Table 1

Characteristics of the children and interventions aspects of the trials that used vestibular rehabilitation exercises programs to improve the postural control, balance and gait of children with SNHL.

Author	Country	Motor Skill	Design	Characteristics of volunteers	Number of volunteers		Characteristics of Interventions	
					CG	IG	CG	IG
Rine et al. [31]	USA	Postural control	RCT	Children with SNHL (moderate to profound hearing loss degrees), both sexes, age range between 3 and 8 years old and bilateral vestibular dysfunction.	11	10	Intervention after the study.	30 min sessions, three times/week of exercise sessions included 10 min of activities in three of four categories: eye hand coordination, general coordination activities, visual motor training, and balance training, for 12 weeks.
Ebrahimi et al. [32]	Iran	Postural control	RCT	Children with SNHL (hearing loss degrees > 90 dB), both sexes, age range between 7 and 12 years old and bilateral vestibular dysfunction.	12	12	Intervention after the study.	45 min sessions, three times/week of a progressive exercise program that included adaptation, eye-head coordination, and substitution exercises to improve gaze and postural stability, for 8 weeks.
Effgen [33]	USA	Balance	RCT	Children with SNHL (hearing loss degrees > 75 dB), both sexes and age range between 7 and 11 years old.	24	25	Normal classroom, activity (Free play).	15 min sessions, of static balance exercises program was conducted daily for 10 consecutive school day.
Rajendran et al. [34]	India	Balance	RCT	Children with SNHL (hearing loss degrees > 90 dB), with or without cochlear implants, both sexes and age range between 6 and 11 years old.	10	11	No intervention.	45 min sessions, three times/week, which included activities in three categories Eye-hand coordination, balance retraining, fundamental motor skills, for 6 weeks.
Soori et al. [35]	Iran	Balance	RTC	Girls with SNHL (hearing loss degrees > 61 dB), with age range between 8 and 11 years old.	10	10	No intervention	60 min sessions, three times/weeks with a perceptual-motor training for 8 weeks.
Demirel [36]	Turkey	Gait	Quasi-randomized trial	Children with SNHL, both sexes and age range between 6 and 9 years old.	09	09	No intervention.	50–75 min sessions, twice times/week: with a special movement-training program, for 10 weeks.

RCT: Randomized controlled trial; SNHL: Sensorineural hearing loss; dB: decibels; CG: Control group; IG: Intervention group.

Table 2

Characteristics of the methodological aspects and conclusions of the trials that used vestibular rehabilitation exercises programs to improve the postural control, balance and gait of children with SNHL.

Author	Country	Motor Skill	Outcome measures	Instruments used for the assessment	Control group		Intervention group		Conclusions
					Pre	Post	Pre	Post	
Rine et al. [31]	USA	Postural control	Body sway	Computerized dynamic posturography by the sensory conditions testing	PSCT-3: 0.18 Somatosensory: 1.43 Vision: 0.31	–	PSCT-3: 2.20 Somatosensory: 0.59 Vision: 0.78	–	Exercise intervention focused on substitution strategies may halt the progression of motor development delay and enhance postural control abilities of children with SNHL and vestibular impairment.
Ebrahimi et al. [32]	Iran	Postural control	Body sway	Computerized dynamic posturography by the sensory organization test	AP: 44.2 ± 9.25 ML: 47.0 ± 8.68	AP: 44.7 ± 9.11 ML: 47.7 ± 9.13	AP: 39.6 ± 6.00 ML: 46.6 ± 6.62	AP: 53.9 ± 6.99 ML: 60.2 ± 6.69	Vestibular rehabilitation therapy focused on adaptation and substitution has shown to improve vestibular function and postural control ability in children with sensorineural hearing loss and vestibular dysfunction.
Effgen [33]	USA	Balance	Body sway	Force platform		AP (EO): 0.00 ± 0.37 AP (EC): 0.08 ± 0.81 ML (EO): -0.06 ± 0.31 ML (EC): -0.02 ± 0.74 RL (EO): 6.68 LL (EO): 8.10		AP (EO): 0.42 ± 2.38 AP (EC): 0.14 ± 0.50 ML (EO): 0.24 ± 1.32 ML (EC): 0.06 ± 0.54 RL (EO): 7.72 LL (EO): 10.2	An exercise program of static balance activities can influence static balance in deaf children by increasing the length of time they can stand on one leg.
Rajendran et al. [34]	India	Balance	Stability limits in the functional reach	Pediatric Reach test (Front)	15.5	16.2	13.5	20.1	A vestibular-specific neuromuscular training program may improve the motor skills, balance and health-related quality of life in children with hearing impairment.
			Stability limits in the functional reach	Pediatric Reach Test (Side)	12.3	13.0	12.0	15.6	
Soori et al. [35]	Iran	Balance	Length time stood on one leg	One leg standing test (Seconds)	EO: 76.8 EC: 20.0	EO: 78.0 EC: 20.3	EO: 75.4 EC: 20.8	EO: 87.1 EC: 24.1	Perceptual-motor training may lead to improvements in coordination and balance of children with hearing impairment.
			Static Balance	Stork Balance test (Seconds)	14.0 ± 7.37	14.6 ± 6.30	14.1 ± 7.63	26.9 ± 7.83	
			Dynamic Balance	Y Balance test	9.07 ± 1.00	9.22 ± 1.12	9.45 ± 12.7	13.9 ± 5.83	
Demirel [36]	Turkey	Gait	Locomotion skills (25 m Walking)	Test of Gross Motor Development	5.45	5.33	5.21	4.33	Exercise and movement training programs for hearing-impaired made positive contributions to the motor skills of these children.

AP: Anterior-posterior sway; ML: Medial-lateral sway; EO: Eyes open; EC: Eyes closed; RL: Right leg; LL: Left leg; PSCT: Posturography sensory conditions testing.

2.3. Participants

The studies were included if the participants were children, clinically diagnosed with SNHL (unilateral or bilateral), belonged to both groups (control and intervention), were aged up to 12 years old, and exhibited no physical problems and cognitive or neurological impairment, except the vestibular dysfunction.

2.4. Interventions

The intervention group were treated with interventions that used balance or postural control exercises, which stimulate vestibular system rehabilitation. The control group involved activities of daily living, recreation, leisure, any other intervention or no intervention.

2.5. Outcomes assessed

The primary outcomes assessed by this systematic review were the postural control and balance and the secondary outcome was the gait.

Included were trials that assessed postural control, according to center of pressure oscillation velocity in the anterior-posterior and/or medial-lateral directions, measured in centimeters per second, using the following instruments: force platform or computerized dynamic posturography. The trials that used the BESS scale (Balance Error Scoring System), a numerical scoring scale, to assess postural control were also included.

Trials that assessed balance using a force platform, or computerized dynamic posturography by the area of center of pressure oscillation, measured in cm^2 , were included. Also included were studies that assessed balance using the following instruments: Pediatric Balance Scale, Pediatric Reach test (front or side), Test of Gross Motor Development, Motor Proficiency test, One Leg Standing test, Peabody Developmental Motor Scales and Movement Assessment Battery for Children (MABC). All of these are clinical tests or scales widely used in clinical practice and by studies that evaluated the balance in children.

Included were studies that evaluated gait with any condition related to walking, such as gait speed, distance between feet, stride length, running or any other means of locomotion. Also included were investigations that assessed gait using the following instruments: Dynamic Gait Index, Timed Up and Go test, by accelerometry, camcorders, photos, and material such as dye to mark the footsteps of children on the floor or paper, or those that used sand for this purpose.

2.6. Data extraction and analysis

The studies data included in this review were extracted and recorded on a standardized form created by the authors. These data were filed independently by both reviewers in Review Manager (RevMan) software, version 5.1.6, for subsequent analysis of the information and discussion of the possible discrepancies. The risk of bias was qualitatively described.

3. Results

3.1. Flow of the trials through the review

A total of 8438 articles were identified according to the search strategies in the nine databases. After duplicate articles were removed, 7107 articles remained which were analyzed one by one by their titles and abstracts. Seventeen of these articles were selected to be read in their entirety. After this analysis, six trials were considered eligible for this systematic review, five randomized [31–35] and one quasi-randomized [36], as shown in Fig. 1, which contains the study extraction flowchart, in line with PRISMA recommendations.

Five of the studies excluded [37–41] used sports or recreational activities as intervention. In two trials [42,43], the control group

consisted of normal-hearing children and two others [44,45] investigated children older than twelve years old and the authors did not stratify the results according to age ranges. One study [46] included children with neurological disorders and another [47] only contained the intervention group.

3.2. Characteristics of the selected trials

All the studies included in this review compared vestibular rehabilitation exercises programs with activities of daily living, recreational activities or no intervention at all. The characteristics of the studies selected, in terms of the characterization of the children, their methodological aspects and conclusions are described in Tables 1 and 2.

3.3. Risk of bias

With respect to randomization of the children, five [31–35] of the six studies were randomized; however, only two [31,34] clearly described how the randomization was conducted. One study [36] did not even report the randomization, but only that the children were divided into intervention and control groups, exhibiting high risk of bias.

Regarding to anonymous allocation, only Rajendran et al. [34] described how this was carried out (by sealed envelopes), while the remaining authors did not report if or how it was performed in their trials, indicating a high risk of bias for these trials.

None of the studies mentioned the blinding of the children and only Rine et al. [31] reported that the evaluators of outcomes did not know which group the children evaluated belonged to (control or intervention), representing high risk of bias for the trials that did not control the blinding of examiners of the outcomes.

In relation to incomplete data or losses, Effgen [33] stated that some of the children in your study were unable to remain in single leg stance (test position) during assessment. Statistical analyzes were conducted without the data of these children, there was no intention-to-treat analysis and, precisely in this variable, the significant improvements in the children were observed, representing a high risk of bias for this trial.

With respect to selective description of outcome, Rine et al. [31] assessed children in six positions, under different sensory conditions (eyes open and closed). However, separate data in each of the six positions were not demonstrated, only collective data of visual, somatosensory and vestibular domains, resulting in the selective description of the outcome in this trial.

Only two trials [31,32] controlled the vestibular function of children with SNHL, which should be controlled since it directly influences the results. Children with SNHL and associated vestibular dysfunction displayed the worst performance in balance and motor skills and this may underestimate the effect size of the interventions, characterizing high risk of bias for studies that did not control the vestibular dysfunction of the children. This can be seen in Figs. 2 and 3, and in Table 3, which depict critical analysis of the risk of bias of the trials and the table of quality of evidence of the GRADE, respectively.

3.4. Participants

The six trials consisted of 153 participants and investigated the effectiveness of the vestibular rehabilitation exercises programs in improving the motor skills of children with SNHL. Two investigated the postural control ($n = 45$), three the balance ($n = 90$) and one the gait ($n = 18$).

3.5. Intervention

The two trials whose aim was to improve the postural control of children with SNHL applied interventions with similar characteristics. Rine et al. [31] used hand-eye coordination exercises, general motor

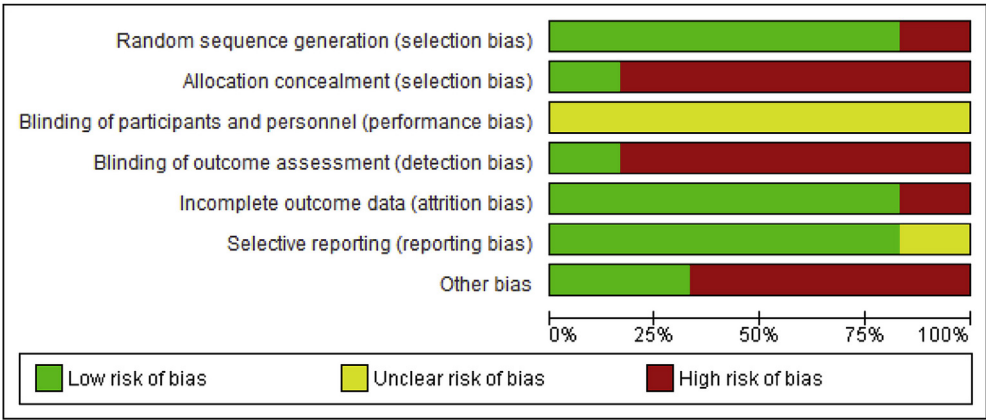


Fig. 2. Risk of bias summary of the included trials.

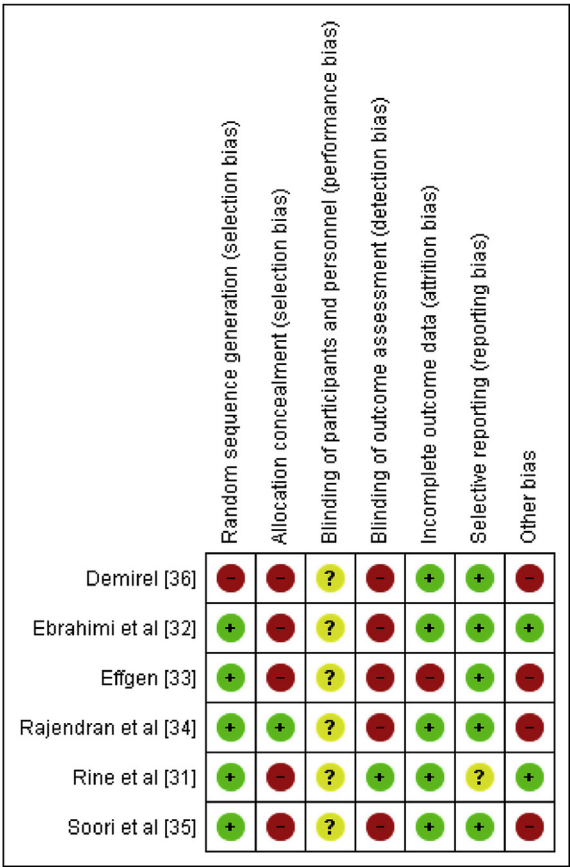


Fig. 3. Risk of bias of each included trial assessed using the Cochrane Risk of Bias Tool.

coordination exercises and visual-motor balance training. Ebrahimi et al. [32] opted for the exercises of adaptation and substitution of hand-eye coordination and training to improve the postural control of the children. In regard to exercise programs, Rine et al. [31] performed 30-min sessions, three sessions were performed during the week and total duration of twelve weeks and Ebrahimi et al. [32] 45-min sessions, three times in the week for eight weeks. In both investigations, the authors observed that the postural control of the children improved significantly after the intervention program (Table 2).

The three trials on balance applied very different interventions. Effgen [33] used static balance exercises with single and double leg stance, Rajendran et al. [34] preferred exercises that were divided into three categories: eye-hand coordination, balance and essential motor

skills, while Soori et al. [35] opted by the motor coordination exercises, agility, jumps and static and dynamic balance exercises in their perceptual-motor training. Regarding to duration of the exercise programs, Effgen [33] held ten 15-min sessions, Rajendran et al. [34] three 45-min sessions on week, for six weeks and Soori et al. [35] used three 60-min sessions on week, during 8 weeks. In all the three trials, the authors reported significant balance improvements of the children after intervention programs. The balance improvements were observed in increased length of stay in unipodal posture (in all the trials); furthermore, Rajendran et al. [34] also reported better vertical balance stability, represented by a decline in the pressure center oscillations of the children and improved functional reach. Soori et al. [35] still observed an improvement in the dynamic balance of the children (Table 2).

Demirel [36] aimed at improving the motor skills of children with SNHL, including gait, using 50-75-min sessions twice a week, for ten weeks. The author found a significant improvement in the motor skills of children, including gait, after the exercise program (Table 2).

None of the studies in this review compared vestibular rehabilitation exercises programs with other interventions for the outcomes analyzed, that is, the control group did not undergo motor intervention simultaneously to the intervention group. In a large number of the studies [33–36], the control group performed activities of daily living, recreational and leisure activities, or no activity whatsoever. Rine et al. [31] and Ebrahimi et al. [32] reported that the control group children were submitted to the same exercise program as the intervention group after the post-test.

3.6. Outcome measures

Postural control was measured by computerized dynamic posturography in both studies. Rine et al. [31] used posturography sensory condition test and Ebrahimi et al. [32] the sensory organization test. Both investigations used the gold standard to evaluate postural control, given that computerized dynamic posturography is capable of providing isolated data from each sensory system (visual, somatosensory and vestibular) exhibiting the main limitations of child, should they exist.

The studies on balance assessed this outcome using different instruments. In your quantitative assessment, Effgen [33] used a force platform, where the number of center of pressure oscillations indicates the stability of the outcome, Rajendran et al. [34] used a sway meter attached to the child's waist. Both studies used the One Leg Standing test, recording the time in seconds that children managed to remain in single leg stance. Rajendran et al. [34] still used the Pediatric Reach test (front and side) to assess the balance stability limits of the children. Soori et al. [35] opted for the Stork Balance test, to evaluate the static balance of the children and the Y Balance test for assess the dynamic

Table 3

Quality of the evidence using the GRADE system (Vestibular rehabilitation exercises programs versus control) to improve the postural control, balance and gait of children with SNHL.

Quality assessment							№ of patients		Quality of evidence	Importance
№ of studies	Study design	Risk of bias	Inconsistency	Indirectness	Imprecision	Other considerations	Vestibular rehabilitation	Control		
Postural control (follow up: mean 10 weeks; assessed with: Computerized Dynamic Posturography)										
02 [31,32]	Randomized controlled trials	serious ^{b,c,d,f}	not serious	serious ⁱ	not serious	none	22	23	⊕⊕○○ LOW	CRITICAL
Balance (follow up: mean 7 weeks; assessed with: Force platform, sway meter and clinical balance tests)										
03 [33–35]	Randomized controlled trials	very serious ^{b,c,d,e,g}	not serious	very serious ^h	not serious	all plausible residual confounding would reduce or improve the demonstrated effect ^g	46	44	⊕○○○ VERY LOW	CRITICAL
Gait (follow up: 10 weeks; assessed with: Test of Gross Motor Development)										
01 [36]	Quasi-randomized trial	very serious ^{a,b,c,d,g}	not serious	not serious	not serious	all plausible residual confounding would reduce or improve the demonstrated effect ^g	09	09	⊕○○○ VERY LOW	CRITICAL

^a There was no random sequence generation.

^b No allocation secrecy.

^c There was no blinding of the children.

^d There was no blinding of the evaluator of outcome.

^e Loss or incomplete data.

^f There was selective reporting of outcome.

^g There was no evaluation of children's vestibular function.

^h Presence of children with cochlear implants.

ⁱ Inclusion of very young children (under seven years old).

balance.

Two trials used the Gross Motor Development test to evaluate the motor skills of children with SNHL, including the gait. However, Demirel [36] demonstrated the gait outcome isolated and Rajendran et al. [34] the same result along with other aspects related to gait and locomotion, which may not specifically represent the gait outcome. Thus, we did not include this study in our gait results.

4. Discussion

Randomized controlled trials with a specific intervention, using vestibular rehabilitation exercises to improve the motor skills of the children with SNHL are scarce in the literature. Nevertheless, the six studies assessed here found that vestibular rehabilitation exercises programs had positive effects on the postural control, balance and gait of these children, thereby improving their motor skills. However, these investigations displayed low or very low methodological quality, for these outcomes, respectively.

The six trials analyzed in this systematic review exhibited important methodological limitations and heterogeneity in the following aspects: session duration, nature of the exercises proposed and total duration of the vestibular rehabilitation program. Moreover, a variety of instruments were used to assess the outcomes and specific and important characteristics of the children were not investigated, since some trials assessed vestibular function and others did not, which precluded meta-analysis of the trial results of the trials included in this systematic review.

Meta-analysis could be conducted between the two trials on postural control, since both used computerized dynamic posturography to assess the outcome, but Rine et al. [31] stratified the variable in their study. Another meta-analysis possibility would be between balance studies that used the One Leg Standing test to assess the static balance; however, Effgen [33] evaluated the right, then the left lower limb and Rajendran et al. [34] only reported that the test was conducted in single leg stance, not specifying whether it was on the child's right, left or preferred leg. We emailed the authors to obtain this information, but received no response, precluding meta-analysis of results in this

systematic review.

Although this is the first systematic review to assess the evidence quality of trials that used vestibular rehabilitation exercises programs to improve postural control, balance and gait of children with SNHL applying the GRADE system. Others systematic reviews observed that these exercises improved the balance, gait and functionality of adults and seniors with vestibular dysfunction [48], presenting quality of evidence from moderate to high [49–52]. Thus, we believe that vestibular rehabilitation exercises programs may also benefit children with SNHL and associated vestibular dysfunction, if they are better planned and controlled in the future trials.

4.1. Cochlear implant-related bias

Another important aspect to consider in future trials is to include children with and without cochlear implants in the same trial, as observed in the study by Rajendran et al. [34]. Only this study included these children, while the remainder did not contain this information nor was it an exclusion criterion, which makes it difficult to know if there were children with cochlear implants in their samples. This information was also not found in the sample characterization of the articles, which is relevant because there is evidence that the children with cochlear implants present a high frequency of vestibular dysfunction [53–57] and four of the six trials of this systematic review did not evaluated the vestibular function of the children, including Rajendran et al. [34] study.

Thus, including children with cochlear implants in the trials and not controlling the vestibular function of the sample may have a confounding effect and underestimate the effect size of the intervention, since these children also exhibit balance and motor development disorders associated with vestibular dysfunctions [57–60]. As such, at the end of the trial, in addition to the confounding effect, it would be difficult to guarantee that the results obtained by the interventions used, would be effective also for children with cochlear implants.

Despite the negative influence on the vestibular system, cochlear implants have shown positive effects, thereby enhancing the auditory and communication skills of implanted children [61–64]. Some studies

observed that children with cochlear implants improved postural stability and balance, after implantation [65–68], most likely because they could hear again, suggesting that auditory input is not neutral in the motor skills [69]. Similar findings were also obtained by other investigations reporting that auditory clues may be able to enhance balance of adults and seniors, using sound amplification devices or cochlear implants [70–74].

This improvement in balance of the children with cochlear implants may be due to the fact that sound signals serve as fixed environmental reference points [75–77], providing individuals with spatial maps of the environment, better space-time orientation and balance for these subjects [77,78].

This condition justifies the need to exclude children with cochlear implants from the trials with children without implants, given that the cochlear implant improve the hearing of these children, who may benefit from this mechanism, thereby producing better results after intervention, overestimating the effect size of the interventions of the trials that included in their samples children with cochlear implants.

As stated above, including children with cochlear implants in the trials analyzed by this review or in the future trials may influence the results of interventions, under- or overestimating the effect size, primarily of investigations that did not control vestibular function in the sample. Unless there is a stratification of the trials results, demonstrating the isolated data of children with and without cochlear implants (a subgroup analysis), which did not occur in the trials analyzed by this systematic review.

4.2. Vestibular dysfunction-related bias

Another important limitation of the trials included in this systematic review was not controlling the vestibular dysfunction of the children, since the fact that children with SNHL exhibit postural control instabilities and changes on balance and gait has been widely associated with vestibular dysfunctions frequently observed in this population. As such, it is indispensable to control vestibular dysfunction in future trials, in order to ensure the external validity of the effects of vestibular rehabilitation exercises programs for children with SNHL with and without vestibular dysfunctions.

One possibility would be to stratify the groups of children according to their vestibular function, since there is evidence that the children with SNHL and associated vestibular dysfunction display the most serious problems in the balance and worse motor performance compared to normal-hearing children [79–82]. In addition, some studies have observed that isolated SNHL has been a triggering factor for balance disorders in children, when compared to normal-hearing children, even in the absence of vestibular dysfunctions [80,82]. Justifying the insertion of children with SNHL without vestibular dysfunctions in rehabilitation programs of balance and motor skills.

Controlling the vestibular dysfunction of children with SNHL, in future trials may guide rehabilitation strategies, such that the positive effects of an adequate intervention can benefit children with severe motor and balance impairment. Not controlling vestibular function, on the other hand, may influence and mask the effect of the intervention, underestimating the effectiveness of vestibular rehabilitation exercises programs, which may have occurred, for example, in the Effgen study [33], where no significant improvements were observed on the balance of children after the intervention, in their quantitative evaluation using a force platform. However, the vestibular dysfunction of the children was not controlled in this trial.

In the aforementioned study [33], the sample could contain children with vestibular dysfunctions, who may have required a longer intervention time than that proposed (15 min). Thus, the effect of intervention may have been underestimated since the vestibular function of the sample was not controlled. Furthermore, the children of this study exhibited greater hearing loss degrees (severe or profound), and several studies have observed a correlation between the extent of hearing loss

and the presence of vestibular dysfunction in children [83–87].

On the other hand, children with SNHL without vestibular dysfunction could respond faster to intervention, requiring a shorter rehabilitation time.

This fact may be associated with the results reported by Demirel [36], where significant improvements on several motor skills were observed after the intervention, but, once again, the vestibular function of the children was not assessed, raising doubts regarding the number of children in the sample without vestibular dysfunction. This intervention may have been longer than necessary to rehabilitate these children (50–75 min); however, these hypotheses regarding the studies of Effgen [33] and Demirel [36] only can be confirmed or rejected by the future trials that evaluated the vestibular system function of the samples.

Regarding evaluation of the vestibular function, De Kegel et al. [79] found that the asymmetry of VEMP (vestibular evoked myogenic potential) could be a good predictor of balance disorders in children with SNHL. Thus, this method of assess of the vestibular function could be one of those chosen by future trials on this topic to evaluate the function of the vestibular system of their samples.

The influences (positive or negative) of cochlear implant and vestibular function on the motor performance of children with SNHL pointed in this systematic review were ignored by the published systematic reviews on this topic. None of the reviews described how these two important characteristics of the children with SNHL could influence the trials results, under or overestimating the effect size of the interventions, demonstrating the importance of this systematic review for the literature and future trials on this topic.

4.3. Limitations and others biases of the trials

Other observations related to the studies included in this review should be mentioned. Sample size was quite small, since a large number used between nine and twelve children in each group. Only Effgen [33] used a larger sample (49 children) and this limitation hinders generalization of the results. Furthermore, none of the authors specified if they used calculations to establish sample size. Another much neglected aspect was anonymous allocation, with only Rajendran et al. [34] detailing how it was conducted, a shortcoming that must be improved in future trials.

Still about the samples, Rine et al. [31] included in their trial very young children, aged between three and eight and a half years old. According to Shumway-Cook and Woollacott [88] the processes that are responsible for resolving multi modal sensory conflict of postural stability are not fully developed before the age of seven years old and suggest that the stability of postural control in children is mature around 7–10 years old, characterizing a selection bias for this trial. Thus, the future trials should categorize age groups if they wish to include children younger than seven years of age, and to observe the results of the interventions in children with postural stability mature or in development, and thus help to guide the clinical practice of these children.

An another aspect that should be better described and detailed on the samples of the future trials is whether the children has unilateral or bilateral hearing loss. Although this systematic review have included trials of children with both hearing loss (unilateral or bilateral), only Rine et al. [31] reported that in their trial all the children had bilateral hearing loss, and the other trials reported only the degrees of hearing loss of the sample. Reporting whether the children has unilateral or bilateral hearing loss is a valuable information, as it assists in understanding on the effectiveness of vestibular rehabilitation exercise programs for the children with different hearing possibilities and contributes to guide the motor rehabilitation in children with SNHL.

Another studies characteristic that needs to be better explained is the exercise used in the interventions. Only Effgen [33] and Demirel [36] provided this informations, while all the others only described the nature of these exercises (coordination, agility, eye-hand exercise and

balance), but without including data on the movements the children performed in the program or the number of series and repetitions, hindering their replication in the clinical practice of the children with SNHL.

Another questionable aspect was that the exercises used were based on the vestibular rehabilitation programs proposed for adults and athletes [31,32,34]. According to the authors, the programs were adapted to the children, but the adaptations were not detailed by any of the studies. With respect to the intensity and difficulty of the exercises, some of the authors reported that the exercises became more difficult over the intervention, but they did not explain how this occurred, or their frequency. Representing a further difficulty for replicating these interventions in rehabilitation of the children with SNHL.

In addition, the studies did not mention whether the children with SNHL used any medication, such as anti-vertiginous drugs, or if they engaged in recreational or sports activities simultaneously to the study, such as physical education, for example, and both factors could affect their motor performance. None of the studies reported whether or not there were adverse effects from the intervention and this information should be included in future trials, since it is relevant, and helps guide the recommendation or not of vestibular rehabilitation exercises programs to treat the motor and balance disorders in children with SNHL.

4.4. Implications for future studies

A limitation of this systematic review was not including theses or dissertations. It is also important to underscore that we changed the title of this review, as stated in the protocol: from “neuromuscular training programs” to “vestibular rehabilitation exercises programs”. The purpose of the change was to reflect the real purpose in the title and not confuse the readers, who could interpret it as being aimed at endurance or sports activities and not therapeutic, as was proposed by this systematic review.

This review observed positive effects of the interventions on the postural control, balance and gait of children with SNHL in trials in which the vestibular rehabilitation exercises programs involved sessions of 30 min or longer and total duration of six or more weeks [31,32,34–36]. This may help guide the indication of exercise-based therapy for children with SNHL and future trials on this issue.

Future trials should also consider the milestones of infant neuro-motor development, an aspect that has been neglected in children with SNHL. There is evidence that motor impairment in this population starts in the first year of life, underscoring motor delay in head control, independent gait and gross motor function [89–93].

Another question that could be addressed in future trials would be to propose an intervention for children with cochlear implants, since, in this extensive review conducted we observed that to date no studies have been carried out to improve or reverse motor and balance disorders in this population. Thus, it is essential that studies consider the specific traits of these children such as whether the implant was uni or bilateral, hearing levels, time since surgery, the child's age, etiology of hearing loss and function of the vestibular system. This information is valuable, since procedures aimed at the motor rehabilitation of children with cochlear implants are currently applied, but still without the necessary theoretical-scientific support.

4.5. Need for advances in integral care and rehabilitation of child with hearing loss

We would also like to emphasize that during this literature survey, evidence from as early as the 1930s found that children with SNHL could present with vestibular disorders [94]. These disorders have been observed decade after decade [95–100] until the present time [81,82,87] and the balance problems since the 1950s [101–106]. Nevertheless, there is still no screening a protocol for these children in terms of rehabilitating these motor disorders and, as observed in the

present updated review, six randomized or quasi-randomized trials proposed motor rehabilitation for the children with SNHL, which is insufficient, given the needs of children with SNHL and its prevalence worldwide.

This demonstrates how rehabilitation with Physiotherapy is still far from children with SNHL, who are in dire need of care, and the extent to which the scientific community must evolve in this topic, in terms of applying the evidence uncovered so many years ago.

Possibly, a challenge in terms of vestibular rehabilitation for this population is that this service are hardly available for free, making it difficult for these children to access quality therapy. Which further reinforces the need to perform further trials on this topic to help prove the effectiveness of vestibular rehabilitation exercise programs to improve the motor skills of children with SNHL. Thus, this treatment could be offered to these children based on high quality evidence and still help implement vestibular rehabilitation in public health services, funded by governmental or public institutions, such as rehabilitation centers and schools aimed at children with SNHL.

There is still evidence that the motor skill deficits affect also adolescents with SNHL [107–109]. As such, we suggest that vestibular rehabilitation exercises programs for this population be planned not only for childhood but for adolescence as well.

We believe that the multiprofessional team (Pediatricians and Pediatricians Otorhinolaryngologists) could act together to assess these children and provide neuromotor and otoneurological diagnoses, respectively, at the time SNHL is diagnosed in neonatal hearing screening programs and refer these children to Physiotherapy assessment and treatment, along with the multidisciplinary team of rehabilitation staffed by Speech and Occupational therapists. What seems to be starting to happen in some countries, such as Belgium, for example [110].

The physiotherapists stand out on the multiprofessional teams for their skills and competencies, marshalling efforts for the common objective of providing comprehensive care to children, with a view to adapting and/or optimizing the motor performance, balance, motor skills and quality of life of children with SNHL.

5. Conclusions and future directions

We concluded that there is promising evidence that the vestibular rehabilitation exercises programs improve the postural control, balance and gait of children with SNHL. However, due to the methodological limitations and low current evidence quality on the topic, the result on the effectiveness of this intervention to improve the postural control, balance and gait of children with SNHL was inconclusive and the trials results analyzed by this systematic review should be interpreted with caution.

Moreover, the adverse effects of vestibular rehabilitation exercises programs were not reported by any of the trials, this makes it difficult to evaluate the relationship between benefits and adverse effects of the interventions. Thus, any estimation presented by this review would be uncertain, inconclusive or insufficient to support a recommendation involving the use of the vestibular rehabilitation exercises to rehabilitate the balance and gait of children with SNHL.

Due to the low quality of evidence observed in this systematic review, we suggest that new trials be proposed on the topic, with better methodological quality to prove the effectiveness of this treatment in the postural control instabilities and on the balance and gait disorders of children with SNHL.

The future trials should control selection bias, the blinding of the outcome evaluators, the sample losses, the selective description of outcome, assess the vestibular system function of the children and use validated and rigorous instruments for the evaluation of outcomes. We also believe still that, in addition to assess the postural control, balance and gait, functional and clinical outcomes should also be investigated. Thus, the next trials could also include the following outcomes: the

functionality, quality of life, running, frequency of falls, vertical jumping and the performance of these children in the sports and recreational practices, aspects that have been few researched or ignored by the trials present in the literature to date.

Future trials could also observe the time required for children to improve their motor skills, the best time duration of sessions, weekly frequency and the characteristics or nature of exercises that produce the best results for the motor rehabilitation of children with SNHL with and without vestibular dysfunctions. Next trials could also investigate the duration time of the effects of these interventions in the children.

Another relevant aspect that could be investigated in next trials on the topic is the satisfaction of the parents and of the child on the interventions, reporting the domains and functional or daily life skills that have been improved in the children after interventions. In addition, the positive and negative points of these interventions, from the perspective of the parents and of the child could be reported. Making the intervention centered on the child's participation, involving the parents in therapy and providing evidence for future therapeutic decision-making on this issue. All these information's will supply theoretical-scientific support to guide the clinical practice and motor rehabilitation of the children with SNHL.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.ijporl.2019.109650>.

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