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Identification of vestibular loss in children with sensorineural hearing loss using the balance subset of the BOT-2 test

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Abstract

Background Vestibular loss in children with sensorineural hearing loss (SNHL) is quite high. Despite the high prevalence of vestibular loss and balance impairment in children with SNHL, they are rarely assessed by clinicians and therefore are commonly undiagnosed. The purpose of the study was to evaluate the balance subset of the Bruininks-Oseretsky Test (BOT-2) as a tool to identify vestibular loss in children with SNHL and to determine its predictive values for vestibular loss.

Methods The study included 210 children allocated into 4 groups: group 1 (control healthy children), group II (children with SNHL but without vestibular loss), group III (children with SNHL and unilateral vestibular loss), and group IV (children with SNHL and bilateral vestibular loss). Caloric test, video head impulse test (vHIT), and cervical vestibular evoked myogenic potential (c-VEMP) test were used for vestibular assessment in children with SNHL and to allocate them accordingly into one of the aforementioned groups. Scores of the balance subset of the BOT-2 were compared among the three groups and compared to the control healthy children.

Results 21.4% of children with SNHL but without vestibular loss have balance deficit revealed by the balance subset of the BOT-2, reflecting its better sensitivity for detecting balance deficit than the physiologic vestibular tests. Children with unilateral or bilateral vestibular loss (groups III and IV) showed a more significant balance deficit than children without vestibular loss (group II). The worst balance score was found in children with bilateral vestibular loss. The positive predictive value of the BOT-2 for peripheral vestibular loss in children with SNHL was 88%.

Conclusion The balance subset of the BOT-2 has particularly good predictive values for vestibular loss in children with SNHL. The test is a simple, easy, fast office test which does not require any costly equipment.

Keywords Sensorineural hearing loss, BOT-2, Vestibular loss

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Background

The prevalence of vestibular loss in children with sensorineural hearing loss (SNHL) is quite high ranging from 20 to 70% [1, 2]. The percentage is highest in children with profound SNHL, where more than 50% of children in this category have vestibular loss and 35% of those children have severe or total vestibular loss [3, 4]. This is commonly attributed to structural damage of the vestibular end organs (saccule, utricle, and semicircular canals). Vestibular assessment typically includes vestibular evoked myogenic potential (VEMP), caloric test, and rotary chair testing [5, 6]. The association between SNHL and vestibular loss can be easily explained by the common embryological development, anatomical location, and the similar physiology of the cochlea and vestibular end organs, which makes vestibular loss the single most common comorbidity of SNHL.

Vestibular system has a major role in motor development. Therefore, vestibular loss associated with congenital SNHL can lead to delayed motor development in infants and children. Delayed sitting, standing, and walking are common among children with congenital SNHL. When those children grow up, the concomitant vestibular loss results in poor balance skills. Inadequate postural control leads to frequent falls, especially during high-level motor activities such as hopping, skipping, or walking on a balance beam. Children with vestibular loss are more vulnerable to traumatic injuries compared to normal children [7]. Therefore, identification of vestibular loss in children with SNHL, assessment of their age-appropriate balance functions, and provision of early rehabilitation are crucial to reduce its negative sequelae.

Despite the high prevalence of vestibular loss in children with SNHL, they are rarely being assessed by clinicians and, therefore are commonly undiagnosed. Pediatric vestibular assessment is challenging and requires high-technology equipment to assess different vestibular end organs, e.g., caloric testing, rotary chair, VEMP, and video head impulse test (vHIT). Equipment required to perform these tests are only available in specialized referral centers. Special and advanced training is required for clinicians to be able to appropriately conduct the tests and correctly interpret their results in young children, who might be uncooperative or unable to follow the test instructions. The challenge is more in children with SNHL due to the additional communication difficulty during testing. These factors limit the utility of the standard vestibular assessment for hearing-impaired children.

The Bruininks-Oseretsky Test of Motor Proficiency (BOTMP) was first published in 1978 [8], and then revised in 2002. The latest edition was published in 2005 as the Bruininks-Oseretsky Test of Motor Proficiency, 2nd edition (BOT-2) [9]. The test evaluates gross and fine motor skills in children between the ages of 4 and 18 years of age. The BOT-2 score has become the most widely used standardized measurement of motor proficiency in children. It is used by clinicians, especially physical and occupational therapists as well as researchers to assess motor skills according to the chronological age. The BOT-2 substantially helps in the diagnosis and management of children with motor impairment [10]. It consists of several subsets, which are designed to evaluate the motor-control skills that are essential for postural stability during standing, walking, or reaching [9]. The test is easy to administer, takes only a few minutes, does not require any high-technology equipment, and can be conducted on the vast majority of children including those with SNHL [11]. Previous studies [12, 13] have shown that children with SNHL associated with vestibular loss had significantly poor scores in the balance subset of the BOT-2 compared to normal healthy children. Moreover, there was a high correlation between the balance score and the results of vestibular testing such as caloric testing, rotary chair, VEMP, and vHIT. Nevertheless, the balance subset of the BOT-2 has not been evaluated as a test to detect vestibular loss in children with SNHL.

In the current study, scores of the balance subset of the BOT-2 were compared between normal healthy children, children with SNHL without vestibular loss, and children with SNHL and vestibular loss based on results of caloric testing, vHIT, and cervical VEMP (c-VEMP) test. The objectives of the study were to evaluate the balance subset of the BOT-2 as a test to identify vestibular loss in children with SNHL and to calculate its predictive values for vestibular loss in children with SNHL. The predictive values were chosen as they are the test's probability of correctly identifying a specific disorder in people who might or might not have the disorder [14]. Therefore, the use of predictive values matches the aim of this study to evaluate the potential use of a balance subset of the BOT-2 to identify vestibular loss in children with SNHL.

The results of this study could provide supportive evidence for the use of the balance subset of the BOT-2 as a reliable and affordable tool to identify vestibular loss in young children who are not fit or capable of performing technology-based vestibular tests or when the equipment are unavailable, regardless of the etiology of the vestibular impairment.

Methods

The study was a prospective one. Children who participated in the current study were allocated into four groups. Group I was the control group and Groups II, III, and IV were the study groups. Group II included children with SNHL without vestibular loss. Group III included children with SNHL and concomitant unilateral vestibular loss, and group IV included children with SNHL and concomitant bilateral vestibular loss. All children of the study groups were recruited from children attending the Audio-Vestibular Unit at Minia University Hospital, Minia, Egypt. The control children were recruited from children of medical staff families and patients' relatives at the Audio-Vestibular Unit. Parents of the participants had been informed in detail about the aims of the study and the procedures. All of them signed a written consent for the participation of their children in the study. The study was approved by the ethical research committee at Minia University.

Rotary chair is not available in our hospital. c-VEMP, vHIT, and caloric testing were used for vestibular assessment in children with SNHL. Hamilton et al. [15] and Bachmann et al. [16] reported that vHIT can be performed accurately in young children as young as 3–4 years. In this study, all children with SNHL could successfully complete both c-VEMP and the vHIT testing. Based on our clinical experience, caloric testing was only performed for children who are 10 years or older. Regarding the vHIT, only the lateral semicircular canal was assessed, as its assessment technique and normative data are more established in children compared to the vertical canals [15, 16]. The criteria for the absence of vestibular loss in group II were bilateral normal c-VEMP responses, normal vestibulo-ocular reflex (VOR) gain of lateral canal measured with vHIT (≥ 0.8 with no corrective saccades), and normal caloric response (i.e., caloric response within normal range and difference between right and left ear responses < 25%). The criteria for unilateral vestibular loss in group III were unilateral absent or low amplitude c-VEMP response (i.e., absent response or asymmetry ratio ≥ 40%) [17], unilateral reduced lateral canal VOR gain in the vHIT < 0.7 with corrective saccades) [15], and unilateral absent or weak caloric response (i.e., absent response or ≥ 25% canal weakness) [18]. The criteria for bilateral vestibular loss in group IV were bilaterally absent c-VEMP, bilateral low gain in vHIT (VOR gain < 0.6 in the plain of the lateral semicircular canal with corrective saccades), and bilaterally absent or bilaterally weak caloric response (< 6° total response from each ear) [18].

Children with ages below 4 years were excluded (below the age range for BOT-2). Also, children with neurological, orthopedic, or cognitive disorder were excluded as well as children with conductive hearing loss or middle ear pathologies as conductive impairment can affect VEMP and caloric responses. One-way analysis of variance (ANOVA), Bonferroni post hoc tests, and Fisher exact tests were performed to compare among the groups and between each two groups.

Children in the control and study group were subjected to the following:

1) History taking including full medical (prenatal, perinatal, and postnatal), audiological history, and family history of hearing loss or balance disorders
2) Otoscopic examination
3) Audiological evaluation in the form of:

   - Immittancemetry including tympanometry and acoustic reflex testing at frequencies 0.5, 1, 2 and 4 KHz using GSI TympStar Pro tympanometer.

   - Conditioned play audiometry or conventional audiometry according to the age and reliability of the children to assess the hearing using Madsen Astera audiometer and sound treated booth Amplisilence. Air conduction thresholds were measured in frequencies 0.25, 0.5, 1, 2, 4 and 8 kHz and bone conduction threshold were measured in frequencies 0.5,1, 2 and 4 kHz.

4) Cervical vestibular-evoked myogenic potential testing (c-VEMP) was performed (only for the study groups) using Interacoustics, Eclipse EP25, which is a two-channel evoked potential system with OtoAccess software. The children were tested in the sitting position. Electromyographic (EMG) activity was recorded ipsilaterally from the middle of the sternocleidomastoid muscle using a surface electrode (the active electrode), with the reference electrode on the upper edge of the sternum and a ground electrode on the forehead. Care was taken to place the active electrodes symmetrically on the right and left sternocleidomastoid muscles. During each recording session, children were instructed and assisted to rotate their heads towards the contralateral side from the tested ear to keep the sternocleidomastoid muscle contracted.

500 Hz Tone burst stimuli with 1 ms rise and fall time and 2 ms plateau were used. They were presented at a rate of 5.1 cycles per second through (Telephonics 296D200-2) headphones at 95 dB nHL. The EMG signal was amplified (5000 times), bandpass filtered (30–1500 Hz), and averaged after 100–200 sweeps. The analysis window started 30 ms before stimulus onset and ended 70 ms after stimulus onset. Each ear was stimulated separately and the first ear to be tested was randomly selected. To minimize the effect of variable tonic activity of the sternocleidomastoid muscle on c-VEMP and to ensure equal muscle contraction on both sides, the evoked potential system does not start data acquisition unless the root mean square EMG activity was between 50 and 200 μV. Data acquisition was rejected when root mean square EMG activity was below 50 μV or above 200 μV. The level of root mean square EMG activity was monitored and displayed on the computer screen, allowing the examiner or an assistant to give feedback to the child and assist him to increase or decrease muscle contraction and maintain constant muscle tension.

The measurement obtained for c-VEMP was the peak amplitude difference between the first positive peak (P1) and the first negative peak (N1). The P1–N1 amplitude asymmetry ratio (AR) was computed using the following equation: (larger amplitude – smaller amplitude)/ (larger amplitude + smaller amplitude) × 100.

5) vHIT testing was conducted (only for the study groups) using the ICS Impulse system (GN Otometrics). This system consists of a lightweight goggle with an integrated high-speed camera (250 Hz) fixed to record responses from the right eye and triaxial gyroscopes enabling immediate recording of head and eye movements to assess the VOR gain. To adapt the vHIT system for use with young children with small head size, Bachmann and his collaborators [16] have suggested some modifications which have been used in this study. Children were seated in a standard, fixed-height chair 1 m from a visual target (a sticker) on the wall at eye level. The ICS Impulse system goggles were placed on the children's faces and firmly secured with the elastic band around the back of the head to prevent goggle slippage and subsequent inaccurate gain calculations. A 10-cm gauze was placed inside the elastic band for additional tightness. In addition, a kid's step stool was used to keep the children seated upright and to help stabilize the body during head movements. To ensure proper pupil tracking, its image was centralized in the region of interest box and the crosshair was centered on the pupil. After calibration, children were instructed to maintain focus on the sticker. The children's head was rotated by the examiner using low amplitude yet high-velocity head impulses to obtain VOR responses from the left and right lateral semicircular canals. During testing, each child was asked to answer a few questions about the colorful sticker to maintain his gaze toward the sticker. When attention began to be lost, a new sticker was used. Head impulses were manually delivered by the examiner with unpredictable timing and direction until the gain values of 20 acceptable impulses were obtained.
6) Caloric test was performed (only for the study group) using Visual Eyes 525 Micromedcial Interacoustics VNG equipment and Aqua Stim Micromedical Interacoustics water irrigator. It was performed for older children who could tolerate and were cooperative during the test. The child lied in the supine position with the head tilted 30° forward to make the lateral canal vertical. Four caloric irrigations were performed consecutively. Water was introduced into the ear canal on one side at temperatures of 44 °C then to the other ear. Afterwards same was repeated with 30 °C water. A minimal interval between the successive irrigations was 5 min. The water was irrigated at a rate of 250 mL/30 s, and nystagmus peak slow phase velocity (SPV) was automatically calculated, while the head was in a central position and the infra-red video Goggle was covered. The child was distracted by engaging him/her in conversation or a suitable mental task. Canal weakness was computed according to the following equation: stronger ear peak SPV (sum of cool and warm stimulation)– weaker ear peak SPV)/ (sum of peak SPV of the four irrigations) × 100.
7) Balance subtest of BOT-2:

The BOT-2 was performed in a room free of distractions. The children were instructed to perform the 9 items of the test. Table 1 shows these 9 test items. Details of performing the test and the order are as follows:

Test item (1): standing with feet apart on a line while looking at a target on the wall for 10 s. The number of seconds that the child can maintain his/her position is counted as the raw score (from 0 to 10 s).
Test item (2): walking forward 6 steps on a line on the floor both hands are on the hips. If the child placed one foot or both feet completely off the line before completing 6 steps, the test is stopped. The number of successful steps was recorded as the raw score (from 0 to 6 steps).
Test item (3): standing on the preferred foot on a line on the floor while looking at a target on the wall. Both hands are on the hips, and the other (not preferred) leg is flexed at the knee. The raw score is equal to the number of seconds (from 0 to 10) during which the child maintains this position up to a maximum of 10 s.
Test item (4): standing with feet apart on a line with eyes closed for 10 s. The raw score is the number of seconds the child can maintain his/her position (from 0 to 10).
Test item (5): walking forward 6 steps on a line on the floor with a heel-to-toe gait. Both hands are on the hips. A step is incorrect if one foot or both feet were placed completely off the line, and the toe of the rear foot failed

Table 1 The balance subset set of the BOT-2 test

Test item	Maximum row score
Standing with feet apart with eye opened	10 s
Standing with feet apart with eye closed	10 s
Standing on one leg with eye opened	10 s
Standing on one leg with eye closed	10 s
Walking for 6 steps on a line on the floor	6 steps
Walking forward 6 steps on a line with heel to-toe gait	6 steps
Standing on a balance beam with heel to-toe	10 s
Standing on one leg on a balance beam with eye opened	10 s
Standing on one leg on a balance beam with eye closed	10 s

to touch the heel of the front foot. The raw score is the number of successful steps (from 0 to 6 steps).

Test item (6): standing on the preferred foot on a line on the floor with eyes closed. Both hands are on the hips, and the other (not preferred) leg is flexed at the knee. The raw score is the number of seconds (from 0 to 10) during which the child maintains this position up to a maximum of 10 s.

Test item (7): standing on the preferred foot on a balance beam (100 cm length, 5 cm height, 10 cm width) with eyes open for 10 s. The raw score is the number of seconds (from 0 to 10) during which the child maintains this position up to a maximum of 10 s.

Test item (8): standing on the balance beam with a heel-to-toe for 10 s. The raw score is the number of seconds (from 0 to 10) during which the child maintains this position up to a maximum of 10 s.

Test item (9): standing on the preferred foot on the balance beam with eyes closed for 10 s. The raw score is the number of seconds (from 0 to 10) during which the child maintains this position up to a maximum of 10 s.

The test items (3, 6, 7, and 9) are stopped before 10 s if the child's free leg touches the floor, drops the free leg below a 45° angle, hooks the free leg behind the supporting leg, or shifts the supporting foot out of place. For each test item, a second trial was conducted only if the child could not achieve the maximum score on the first trial, and in such cases, the better score was used for scoring.

The raw score (i.e., seconds or number of steps) in each test item is converted to an equivalent point score. The Total Point Score is computed as the sum of the equivalent point scores of all the nine items. The Total Point Score is converted into the Scale Score according to special tables using age and gender-specific normative data [9]. According to the Scale Score, the child's performance is categorized into one of the following categorical descriptions based on the normative data: (1) average, (2) below average, (3) well below average, (4) above average, and (5) Well above average. Table 2 shows the results of the test of one of the normal control children.

Calculation of the predictive values

The predictive values were calculated as a percentage according to the following formulae:

$$\begin{aligned}&\text{Positive predictive value}\\&= \text{True positive / (True positive + False positive).}\\&= \text{children with SNHL with abnormal BOT} - 2\text{ scores (below average or well below average) and vestibular loss according to the c}\\&-\text{VEMP, vHIT, and caloric testing / children with SNHL, abnormal BOT} - 2\text{ scores and vestibular loss}\\&+\text{children with SNHL and abnormal BOT} - 2\text{ scores but without vestibular loss.}\end{aligned}$$

$$\begin{aligned}&\text{Negative predictive value}\\&= \text{True negative / (True negative + False negative).}\\&= \text{children with SNHL and normal BOT} - 2\text{ score (average or above average) and without vestibular loss according to } c\\&-\text{VEMP, vHIT, and caloric testing / (children with SNHL, normal BOT} - 2\text{ score and without vestibular loss}\\&+\text{children with SNHL, normal BOT} - 2\text{ score but with vestibular loss.)}\end{aligned}$$

Table 2 Scores of the balance subset of the BOT-2 in a normal 7-year-old boy

Test item	Raw score	Equivalent point score
1) Standing feet apart on a line eyes open	10 s	4
2) Walking forward on a line	6 s	4
3) Standing on one leg on a line eyes open	10 s	4
4) Standing with feet apart on a line eyes closed	10 s	4
5) Walking forward heal to toe on a line	6 steps	4
6) Standing on one leg on a line eyes closed	7.8 s	3
7) Standing on one leg on a balance beam eyes open	10 s	4
8) Standing heal to toe on a balance beam	10 s	4
9) Standing on one leg on balance beam eyes closed	2.5 s	1
Total point score	32	
Scale score	15	
Categorical description	Average	

Results

Table 3 shows the demographic data of children who participated in this study and the degree of hearing loss in the study groups. The control group included 130 children. They were normal healthy children with bilateral normal hearing sensitivity and normal developmental history. Their age ranged from 4 to 18 years with a mean of 9.5 years and an SD of 3.3 years. They were 83 boys and 47 girls. Group II included 28 children with bilateral SNHL without vestibular loss. The age of children in this group ranged from 6.8 to 17 years with a mean of 11 years and SD of 3.1 years. They were 16 boys and 12 girls. Group III included 20 children with bilateral SNHL and unilateral vestibular loss. The age of children in this group ranged from 6 to 17 years with a mean of 11.9 years and SD of 2.7 years. There were 13 boys and 7 girls. Group IV included 32 children with bilateral SNHL and bilateral vestibular loss. The age of children in this group ranged from 5 to 16 years with a mean of 10 years and SD of 3.2 years. They were 15 boys and 17 girls. Most of the subjects in the study groups III and IV were children with bilateral severe to profound SNHL who were on the waiting list for cochlear implantation (CI) or already had received unilateral CI, as the prevalence of vestibular loss is quite high in this population. The role of our institute is to provide only unilateral cochlear implantation due to limited resources.

Table 4 shows the results of each item of the balance subset of the BOT-2, the total point score, and the scale score of the control and study groups. Table 5 shows the categorical description of the test in those children. Statistically significant differences among the participating groups were found. Overall, the lowest scores

Table 3 Demographic data of children participated in the study and the degree of hearing loss in the study groups

	Group I	Group II	Group III	Group IV
Number	130	28	20	32
Male	83 (63.8%)	16 (57.1%)	13 (65%)	15 (47%)
Female	47 (36.2%)	12 (42.9%)	7 (35%)	17 (53%)
Age (years)				
Mean	9.5	11	11.9	10
SD	3.3	3.1	2.7	3.2
Minimum	4	6.8	6	5
Maximum	18	17	17	16
Degree of hearing loss in the study groups		Bilateral moderately severe (# = 6) Bilateral severe (# = 9) Right profound, and Left severe (# = 1) Bilateral severe to profound (# = 3) Bilateral profound (# = 9)	Bilateral moderately severe (# = 1) Bilateral profound (# = 19)	Bilateral moderately severe (# = 1) Right profound, and Left severe (# = 1) Bilateral profound (# = 30)

Table 4 One-way ANOVA to compare the control and the study groups as regards the scores obtained at different items of the balance subset of the BOT-2

Test item	Mean ± SD				*F* value	*P* value
	Group I	Group II	Group III	Group IV		
1) Standing with feet apart on a line-eyes open	4 ± 0	4 ± 0	4 ± 0	3.9 ± 0.3	5.918	0.001
2) Walking forward on a line	4 ± 0	4 ± 0	4 ± 0	3.5 ± 1.1	10.594	< 0.001
3) Standing on one leg on a line-eyes open	3.6 ± 0.7	3.4 ± 0.9	2.9 ± 0.9	2.3 ± 1.6	19.748	< 0.001
4) Standing with feet apart on a line-eyes closed	4 ± 0.2	4 ± 0.2	4 ± 0.2	3.8 ± 0.6	4.811	0.003
5) Walking forward heal to toe on a line	3.7 ± 0.8	4 ± 0.2	3.7 ± 0.7	2.3 ± 1.5	27.089	< 0.001
6) Standing on one leg on a line-eyes closed	2.9 ± 0.9	2.6 ± 1	1.8 ± 0.6	0.9 ± 0.8	39.147	< .001
7) Standing on one leg on a balance beam-eyes open	3.7 ± 0.7	3.5 ± 0.8	2.7 ± 0.8	2.2 ± 1.8	24.215	< 0.001
8) Standing heal to toe on a balance beam	3.9 ± 0.3	4 ± 0.2	3.4 ± 0.8	2.9 ± 1.5	19.240	< 0.001
9) Standing on one leg on a balance beam-eyes closed	3.3 ± 1.3	2.7 ± 1.4	1.4 ± 0.5	1 ± 1	40.273	< 0.001
Total point score	33.1 ± 3.1	32 ± 3.4	27.8 ± 2.5	22.7 ± 2.7	47.361	< 0.001
Scale score	17.3 ± 3.7	13.8 ± 4.6	7.9 ± 2.1	7.2 ± 3.1	89.909	< 0.001

Table 5 Comparisons among the control group and study groups as regards the categorical description of the balance subset of BOT-2

	Well below average	Below average	Average	Above average	Well above average	Total
Group I	0 (0%)	0 (0%)	95 (73.08%)	34 (26.15%)	1 (0.77%)	130
Group II	0 (0%)	6 (21.43%)	16 (57.14%)	6 (21.43%)	0 (0%)	28
Group III	2 (10%)	16 (80%)	2 (10%)	0 (0%)	0 (0%)	20
Group IV	14 (43.75%)	12 (37.5%)	6 (18.75%)	0 (0%)	0 (0%)	32
Total						210

Group I: The control group

Group II: children with SNHL without vestibular loss

Group III: children with SNHL and unilateral vestibular loss

Group VI: children with SNHL and bilateral vestibular loss

and the worst category (both reflect more balance deficit) were found in children with bilateral vestibular loss (group IV). The differences between the groups are described as follows:

Comparison between the control children (group I) and children with SNHL without vestibular loss (group II)

There was no statistically significant difference between the control children and SNHL children without vestibular loss in all test items and in the total point score. However, SNHL children had statistically significantly lower scale scores than the scale scores of the control children (p value = < 0.001). The mean and SD of the scale score for the children with SNHL were 13.8 ± 4.6 compared to 17.3 ± 3.7 for control children. Categorically, the Fisher exact test revealed a statistically significant difference between the two groups, whereas 21.4% of the SNHL children without vestibular loss were below average while none of the control group was below average (p value = 0.000). These results reflect that more than a fifth of children with SNHL without vestibular loss have a balance deficit, which can be detected by the balance subset of the BOT-2.

Comparison between the control children (group I) and children with SNHL and vestibular loss (groups III and IV)

Compared to the control children, children with SNHL and unilateral vestibular loss (group III) had reduced scores in multiple items of the balance section of BOT-2. The total point scores and scale scores were statistically lower in group III than in the control group. The mean and SD of the total point score in SNHL children with unilateral vestibular loss were 27.8 ± 2.5 compared to 33.1 ± 3.1 for control children (p value = < 0.001) and mean and SD of the scale score in children with SNHL and unilateral vestibular loss were 7.9 ± 2.1 compared to 17.3 ± 3.7 for control children (p value = < 0.001). The relatively easy tasks are standing feet apart on a line with eyes open (test item 1), walking forward on a line (test item 2), standing feet apart on a line with eyes closed (test item 4), and walking forward heal to toe on a line (test item 5) did not show any statistically significant differences between the control children and children in group III.

Performance of children with SNHL and bilateral vestibular loss (group IV) was poor compared to children in group III, whereas their scores were substantially and statistically lower than the scores of control children in all test items, and in the total point score and scale score. The mean and SD of the total point score in SNHL children with bilateral vestibular loss were 22.7 ± 2.7 compared to 33.1 ± 3.1 for control children (p value = < 0.001) and mean and SD of the scale score in SNHL children with bilateral vestibular loss were 7.2 ± 3.1 compared to 17.3 ± 3.7 for control children (p value = < 0.001).

Comparison between children with SNHL without a vestibular loss (group II) and, children with SNHL with vestibular loss (groups III, and IV)

Compared to children with SNHL without vestibular loss, children with SNHL and unilateral vestibular loss had no statistically significant difference in their scores in the test items 1 to 5 (standing feet apart on a line with eye open, walking forward on a line, standing on one leg on a line with eye open, standing feet apart on a line with eye closed, and walking forward heal to toe on a line). In the other test items, total point score, and scale score, children in group III had statistically significantly lower scores. The scores were least with more statistically significant difference in test item number 9 (standing on one leg on the balance beam with eyes closed), Total point score and the scale scores were 27.8 ± 2.5, and 7.9 ± 2.1 for children with SNHL and unilateral vestibular loss compared to 32 ± 3.4, and 13.8 ± 4.6 for children with SNHL without vestibular loss (P values = 0.001, 0.014, and < 0.001 respectively). Categorically, the Fisher exact test revealed a statistically significant difference between the two groups whereas only 21.4% of children in group II were below average, and 80% of children in group III were below average ($p=0.0000$).

As expected, the performance of children in group IV (SNHL with bilateral vestibular loss) was worse than children with SNHL and unilateral vestibular loss (group III), whereas children in group IV had statistically significantly lower scores than children in group II in all test items, the total point score and the scale score.

Comparison between children with SNHL and unilateral vestibular loss (group III) and children with SNHL and bilateral vestibular loss (group IV)

The performance of children in group IV was worse than the performance of children in group III. Although there was no statistically significant difference between scores of children in group III in some test items (standing on one leg on a line with eyes open, standing on one leg on a balance beam with eye open and eye closed, standing to heal to toe on a balance beam and in the scale score), their score showed a statistically significant difference in the other test items and total point score. Moreover, children in group IV were worse in the categorical description whereas 43.75% of these children were in the well below average category compared to only 10% of children in group III ($p=0.000$).

Prediction of vestibular loss in children with SNHL using the BOT-2

The predictive values of the balance subset of the BOT-2 test for vestibular loss in children with SNHL were calculated according to the formulae mentioned in the methodology section based on the data in Table 6. The positive predictive value was 88% and the negative predictive value was 73%.

Table 6 Predictive values of the BOT-2 test for vestibular loss in children with SNHL children

	Positive vestibular impairment	Negative vestibular impairment	Total
Abnormal BOT-2 test (Below or way below average)	True positive = 44	False positive = 6	50
Normal BOT-2 test (Average or better)	False negative = 8	True negative = 22	30
Total	52	28	80
Positive predictive value	44/44 + 6 = 88%		
Negative predictive value	22/22 + 8 = 73%		

Group I: The control group

Group II: children with SNHL without vestibular loss

Group III: children with SNHL and unilateral vestibular loss

Group VI: children with SNHL and bilateral vestibular loss

Discussion

The current study was designed to evaluate the balance subset of the BOT-2 as a test to predict the presence of a vestibular loss in children with SNHL and to determine its predictive values. After vestibular assessment using c-VEMP, vHIT, and caloric testing, children with SNHL were categorized into three groups: children with SNHL without vestibular loss (group II), children with SNHL and unilateral vestibular loss (group III), and children with SNHL and bilateral vestibular loss (group IV). Results of the balance subset of the BOT-2 were compared among the three groups and compared to the control healthy children (group I).

Balance subtest of BOT-2 in children with SNHL without vestibular loss: more than a fifth of children with SNHL without vestibular loss have balance deficit

There was a statistically significant difference in the scale score between the control children and children with SNHL without vestibular loss, whereas children with SNHL without vestibular loss had a mean scale score of 13.8 compared to 17.3 for the control. Categorically, the Fisher exact test revealed a statistically significant difference between the two groups, whereas 21.4% of children with SNHL without vestibular loss were below average reflecting balance deficit in those children despite the normal vestibular end organ function measured by c-VEMP, vHIT, and caloric testing. There might be subtle vestibular impairment in those children not detected by these tests. Such vestibular deficit can be revealed by the balance subset of the BOT-2, reflecting its better sensitivity for detecting balance deficit than the physiologic vestibular tests.

Another explanation of these findings is the possible role of acoustic and auditory cues in balance and postural control. Children with SNHL without vestibular loss lack this auditory input because of impaired hearing. This explanation is supported by the findings of Sokolov et al. [13], who have reported significantly lower scores of the balance subset of BOT-2 in children with unilateral deafness despite the presence of normal vestibular function. It is highly recommended to assess balance in children with SNHL even if the vestibular tests revealed normal results.

Balance subtest of BOT-2 in children with SNHL and vestibular loss

85% of children with SNHL and vestibular loss have a balance deficit

Results of the balance subset of the BOT-2 in the current study disclosed a significant balance deficit in children with SNHL and unilateral or bilateral vestibular loss. The study revealed that the majority of children with SNHL and unilateral or bilateral vestibular loss had balance deficits. Only 15.3% of these children were within the average category of the balance subset of the BOT-2 and the rest of them were either within the below-average or well below-average category. The balance deficit as revealed by the balance subset of the BOT-2 in children with SNHL and vestibular loss was reported in other studies [12, 13]. Considering the high percentage of children with SNHL who have balance deficit, it is highly recommended to include balance assessment as a routine in the diagnostic workup of children with SNHL as such balance deficit is usually inapparent except in challenging motor activities. Results of the current study showed a very good predictive value of the balance subset of the BOT-2 for vestibular loss in children with SNHL. The test overcomes the barriers that are encountered by clinicians in assessing vestibular functions in young children as the test is simple, easy, reliable, fast, and costless. In addition, it is well tolerated and can be properly conducted on the vast majority of children with SNHL.

Children with bilateral vestibular loss performed worse than children with unilateral vestibular loss

Results of the balance subset of the BOT-2 in the current study revealed that the balance deficit was more marked in children with bilateral vestibular loss than in children with unilateral vestibular loss. While children with unilateral vestibular loss could normally perform the easy BOT-2 items, children with bilateral vestibular loss could not normally perform any item. In addition, children with bilateral vestibular loss had significantly lower scores in some test items and total point scores than the children with unilateral vestibular loss. Categorically, 43.75% of children with bilateral vestibular loss were in the well below-average category compared to only 10% of children with unilateral vestibular loss. The results of the current study are quite consistent with published studies [12, 13], which reported that children with bilateral vestibular loss had significantly lower scores of the balance subset of the BOT-2 than children with unilateral loss. Results of the current study indicate that the balance subset of the BOT-2 is not only a sensitive test to detect balance deficit and vestibular loss in children with SNHL, but it can reflect the severity and the magnitude of the balance impairment through its scores.

Prediction of vestibular loss in children with SNHL using balance subset of the BOT-2

Oyewumi et al. [12] used the balance subset of the BOT-2 to screen for vestibular loss in children with SNHL and cochlear implants. They also measured the sensitivity and specificity of the balance subset of the BOT-2 for vestibular loss in children with SNHL. The test sensitivity was 90% and specificity was 84%. However, Oyewumi et al. [12] included only children with bilateral total vestibular loss defined as (1) bilateral absent caloric response to ice water, and low gain of the vestibular ocular reflex on rotary chair testing or vHIT, as well as (2) bilateral absence of a saccular response as revealed by the absent c-VEMP responses. The current study included children with SNHL, unilateral, and bilateral vestibular loss. The study also included children with a less severe degree of vestibular loss, which is defined as reduced amplitude or absent c-VEMP, low gain in the lateral semicircular canal VOR in vHIT, and weak or absent caloric response. Moreover, the predictive values were chosen over the sensitivity and specificity as sensitivity and specificity are test's probability of correctly identifying the disorder solely from among people who are known to have a condition (sensitivity) or not to have the disorder (specificity). On the other hand, the predictive values are test's probability of correctly identifying the disorder among people who might or might not have the disorder [14]. Therefore, the predictive values suit the aim of the current study to evaluate the balance subset of the BOT-2 to reliably identify vestibular loss in children with SNHL in an efficient non-instrumental way.

Despite the methodological difference, there is quite a consistency between the current study results and the results of Oyewuni et al. [12] as regards the validity of the balance subset of the BOT-2 to predict or detect vestibular loss in children with SNHL. The positive predictive value calculated in the current study was 88% reflecting that the abnormal score of the balance subset of the BOT-2 correctly identified the presence of vestibular loss in children with SNHL by 88% probability. Considering the high prevalence of vestibular loss among children with SNHL and considering the advantages of the BOT-2 as a simple, easy office test that requires just a few minutes, the balance subset of BOT-2 would be extremely valuable clinical tool with high positive predictive value to correctly identify common and important comorbidity such as the vestibular loss in children with SNHL. On the other hand, the calculated negative predictive value was much lower than the positive predictive value (73% for the negative predictive value and 88% for the positive predictive value). The results demonstrate that the balance subset of the BOT-2 is a very good positive test. When positive (abnormal), it correctly identifies vestibular loss in children with SNHL with a very good probability. When negative (normal), it excludes the presence of vestibular impairment by only a 73% probability.

Conclusion

Results of the current study demonstrate particularly good predictive values of the balance section of the BOT-2 to correctly identify vestibular loss in children with SNHL. The test is simple, well tolerated, and easily conducted on the vast majority of children, and most importantly being costless, in addition, administration of the nine items of the test and calculating the scale score only takes a few minutes. It is strongly recommended to apply the balance subset of the BOT-2 for all children with SNHL to identify possible vestibular loss associated with the SNHL and to monitor the balance improvement with vestibular-balance rehabilitation therapy. Furthermore, we propose to use the test to identify vestibular loss in children due to pathologies other than SNHL.

Limitations of the study

The BOT-2 test is limited only to children older than 4 years. Other limitations of this study are being conducted in only one center, and vHIT of vertical canals and ocular VEMP were not tested in this study.

Recommendations

We recommend future replication of the same study in multi-center bases with a larger number of subjects.

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Authors' contributions
All authors contributed to the study. ME: study design, statistical analysis, data analysis, writing the manuscript. RG: performing the vestibular tests (video head impulse test, VEMP testing, and Caloric). MH: performing the vestibular tests (video head impulse test, VEMP testing, and caloric). DM: data analysis and reviewing the manuscript. GA: performing the balance subset of the BOT-2 test; AM: data analysis and Writing the manuscript. All authors read and approved the final manuscript.

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Availability of data and materials
The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate
The study was approved by the ethical research committee of Minia University. Parents of the participants had been informed in detail about the aims of the study and the procedures. All of them signed a written consent for their children's participation in the study.

Consent for publication
Not applicable.

Competing interests
Dr Mohamed El badry and Dr Alfarghal Mohamed are co-authors of this study and Associate Editors for the journal. Dr El badry has not involved in handling this manuscript during review process. Dr Mohamed has not involved in handling this manuscript during the submission and review processes. The rest of the authors have no conflict of interest to declare.

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