How sacculo-collic function assessed by cervical vestibular evoked myogenic Potentials correlates with the quality of postural control in hearing impaired children?

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How sacculo-collic function assessed by cervical Vestibular Evoked Myogenic Potentials correlates with the quality of Postural Control in hearing impaired children?

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Abstract

Objective:

Functional integrity of vestibular end organs is essential for gaze stabilization, dynamic visual acuity, postural control and spatial orientation. Some authors hypothesized on the importance of saccules for postural control and motor development in children, including achievements such as standing up and walking. The purpose of this article was to observe how saccular dysfunction assessed by cervical Vestibular Evoked Myogenic Potentials (cVEMPs) correlates with the quality of postural control in non-syndromic deaf children.

Method:

Seventy-six non-syndromic hearing-impaired children were retrospectively included. Sacculo-collic pathway was assessed with cVEMPs elicited in bone conduction. The response was quoted "normal" if a reproducible wave P13-N23 of at least 50uV in amplitude was present, if not, it was quoted "absent". The sample was divided in 3 groups depending on the presence of the sacculo-collic responses: normal bilateral group (Group 1), normal unilateral (Group 2) and absent bilaterally group (Group 3). Motor assessment was achieved with Movement Assessment Battery for Children, second edition (MABC-2). Postural control (PC) was assessed using the dynamic Balance Quest platform. The scores obtained with MABC-2, and the postural parameters recorded on the Balance Quest platform (sway of Centre of Pressure and spectral power index) were analyzed and compared throughout the groups.

Results:

Group 1 (normal bilateral) showed the best scores regarding motor abilities and postural stability within available normative data. Group 3 (absent bilateral) had the lowest motor and postural control skills. A good correlation between the scores obtained by MABC-2 motor test and dynamic posturography (Balance Quest) was observed.

Conclusions:

The presence of at least one sacculo-collic response would predict satisfactory static and dynamic motor and postural control skills in non-syndromic hearing-impaired children. MABC-2 and Dynamic Posturography Balance Quest appears reliable and comparable tools for PC assessment in hearing impaired children. In the light of these results, it appears that in young children candidates for uni- or bilateral CI whose walking is not yet acquired, should receive a vestibular assessment before surgery to avoid the risk of bilateral sacculo-collic function impairment.

Key words: Hearing impaired children, Vestibular impairment, Sacculo-collic function, Postural control, MABC-2, Dynamic Posturography

1. Introduction

Profoundly located in the labyrinthine partition of the inner ear, the vestibular end organs are accelerometers specialized in quantifying head movements at any moment. More specifically, they allow through sacullar and utricular maculae to assess gravitational or linear accelerations during translational movements and through ampullaris cristae to determine active or passive angular head accelerations. Furthermore, through reflexes originating in the vestibular nuclei, they contribute to ensure visual and head stability in movement, which is essential for spatial orientation. Subsequently, permanent sensory and posturomotor adjustments via the vestibulospinal tract are needed to ensure and adopt the most adequate sensory strategy and motor reaction to restore balance at any moment. The physical principle which applies and corresponds to the essential bases of posturography is to keep the projection of the center of the mass within the sustentation polygon [1]. If the vestibular end system is functional and postural control is matured, imbalance and/or falls are avoided even in the case of a challenging sensory condition (e.g. distorted/absent proprioception and/or vision as in Condition 5 and 6 of the Balance Quest platform).

According to Rine et al., in children with vestibular impairment a motor development delay and a variable deficit of the postural control is common [2]. Vertigo and imbalance are often underestimated in the pediatric population, due to limited communication abilities, atypical symptoms, and relatively quick adaptation and vestibular compensation in children [3]. A posturographic study in a small sample of vestibular impaired children compared the symptoms and clinical signs with the postural parameters before and after physical rehabilitation therapy [4]. The authors found in these subjects a progressive improvement of symptoms in correlation with the postural stability amelioration through physical rehabilitation. In the light of these results, they suggested that subjective vestibular symptoms positively correlate with posturographic data. Rine et al. reported that a delay in postural control development in children did not regularly raise suspicion of vestibular dysfunction due to scarce symptoms [5]. It was therefore suggested that vestibular function should be screened in these subjects, and in case of impairment, further postural control abilities should be explored in order to propose the most appropriate therapeutic strategy.

Wiener-Vacher et al. [6], assessed the Vestibulo-Ocular Response (VOR) in 26 healthy children between 6 and 25 months old, using electro-oculographic recordings (EOG) of VOR to both semicircular canal and saccular stimulation, at three different stages of posturomotor control development: i) before, ii) during their first steps and iii) during the first year of independent walking. Semicircular canal VOR was recorded in seated children, through rotatory impulsions on a vertical axis (acceleration and deceleration both at 40 degrees/s^2, separated by a rotation at 60 degrees/s velocity). The otolith VOR were recorded with a specific device, the Off-Vertical Axis Rotation test (OVAR). The authors reported that the semicircular canal VOR was not related to the age or children postural motor development. However, the otolith VOR parameters changed significantly: the modulation of the horizontal component increased, and the modulation of the vertical component decreased, suggesting that otolith function is mostly linked to the walking maturation process. The results suggested that the otolith VOR variations during the first year of walking were in line with an increase in head-trunk stiffness involved in a specific gaze stabilization strategy. This hypothesis is supported by a recent study [7] in a cohort of children (median age 2.3years) affected by bacterial meningitis. Subjects with severe vestibular impairment due to meningitis with no bilateral cVEMPs prior to walking, exhibited a significant delay in postural control development when compared to similar subjects whose onset of meningitis occurred after acquiring the ability to walk. In the light of these findings, it appears that the otolith system is still developing while learning to walk between 12 and 14 months of age, whereas, as previously reported semicircular canal VOR still matures in growing children aged between 6-12 years [8]. Furthermore, Allum et al. [9] emphasized the importance of a remnant sacculo-otolithic function assessed by cVEMPs, in a subject with severe and brutal onset of a bilateral semicircular canal VOR hypofunction whose balance control compensated better than generally in this case. Consequently, sacculo-otolithic system impairment appears to be involved in case of significant development deficits especially through childhood but not only, having a negative impact on posturomotor abilities.

For more than ten years, vestibular function of children suffering from hearing loss has been attentively studied. Vestibular dysfunction most likely occurs in children with sensorineural hearing loss (SNHL) due to embryological and anatomical connection between the cochlea and vestibular end-organs and their shared sensory microstructure and genetics [5].

The prevalence of vestibular dysfunction in children with hearing loss ranges from 20 to 85% [3, 10-11]. This variability could be related to the different underlying pathologies or the degree of the hearing loss [12]

The goal of the present study was to explore the involvement of saccular function in postural capacities in hearing impaired children.

2. Materials and methods

*2.1.*Subjects

Vestibular data (cVEMPs and Dynamic Posturography Balance Quest, Framiral) from 76 hearing impaired children, 28 girls and 48 boys, from 3.9 to 16.7 years old with the mean age of: 9.3 ± 3.4 years old was studied retrospectively. MABC-2 as a clinical assessment tool was also performed in all participants. A non-syndromic hearing loss (NSHL) was the main including criteria. According to the last recommendation of the French society of otorhinolaryngology (https://www.orlfrance.org/wp-content/uploads/2017/06/Consensus_audiometrie_2016.pdf) NSHL was found profound in 54%, severe in 10.5%, medium in 29% and mild in 6.5% of the participants. Seventy-seven percent were idiopathic. NSHL versus 23% with known disorders

(10.6% morphological with abnormality of the inner ear, 5.3% familial, 3.9% infectious of which 1.3% post-meningitis and 2.6% due to cytomegalovirus). The exclusion criteria were acute otitis media with effusion, migraines and equivalencies including benign paroxysmal vertigo, recent orthopedic or brain traumatic injury, psychiatric disorders, uncorrected vision problems, history of recent crisis of vertigo in less than 6 months.

The investigation adhered to the principles of the Declaration of Helsinki and was approved by our Institutional Human Experimentation Committee. Written informed consent was obtained from the children's parents. The experimental procedure was explained to each child and could have been stopped at any time if requested.

2.2. Assessment

2.2.1. Cervical Vestibular Evoked Myogenic Potential (cVEMP)

The last Pure Tone and Speech Audiometry realized in the Audiology Department of our Tertiary Center was used to determinate the profoundness of the Hearing Loss. Otoscopy and tympanometry were verified in each subject before cVEMPs assessment. Sacculo-collic pathway was assessed by cVEMPs elicited in bone conduction (BC) as previously described for children [13] and recommended by international guidelines [14]. As previously communicated, the utricle may also respond to BC stimuli but the presence of cVEMPs indicates predominantly human saccular response function [15-19]. Standard recording electrodes (standard neonatal ECG, Kendall) were placed over the middle of the SCM belly, and the reference electrode was placed over the medial clavicle, earth being placed on the sternum. The children either lay on a clinical table or sat on their parent's lap. Children were requested to raise their heads and turn it to the right or to the left in order to assess respectively the left and right saccular responses. Stimuli were generated by means of Neuro-Audio software using Neurosoft® (Ivanovo, Russia) laboratory interface and a customized amplifier. The stimulations were delivered by applying the bone oscillator on the mastoid 3 cm posterior and 2 cm superior to the external auditory canal. BC calibrated tone bursts characteristics delivered by a B71 bone conductor vibrator (B71, Radioear, New Eagle, PA) to the mastoids were 500 Hz, 4 ms unshaped sine waves of 136-dB peak force level.

During BC stimulation, the activity of the sternocleidomastoid muscle was recorded by electromyography (EMG). The EMG signals were amplified and bandpass filtered between 30 and 3000 Hz; care was taken to ensure similar background activation of the SCM between sides of the neck and trials. All sweeps that don't fall in the required EMG range were automatically rejected. The early wave's latencies P13 and N23 were recorded. The cVEMPs ratio /asymmetry were automatically provided by the Neuro-Audio software. To check the reproducibility, the responses were recorded twice in both sides.

For the purpose of the present study, response was marked "Normal" if a two-peaked reproducible wave P13-N23 of at least 50uV was present at the highest intensity of stimulation in bone conduction at 70dB HL (**Figure 1**). Similarly, if no reproducible cVEMPs was generated or in case of a response inferior to 50uV, the response was marked "Absent". Based on these results, we divided the findings into three groups depending on whether children had normal bilateral saccular responses (Group 1) or exhibited a saccular deficiency unilateral (Group 2) or bilateral (Group3).

2.2.2. Clinical motor function measures of balance

Physiotherapists have developed several clinical tests which allow clinicians to quantify motor delay in school-aged children between 3 and 16 years old [20]. Particularly the Movement Assessment Battery for Children and teenagers, second edition (MABC-2), is used to assess the motor capacities and thus detect motor delay in children from 3 to 16 years old and eleven months [21]. MABC-2 involves eight tests organized in three subsections: manual dexterity, balance and ball skills (aiming and catching skills). Specific tests are selected from each of these subsections depending on the age of the children in order to attribute specific scores. Standardized and percentile scores are calculated on a validated Dutch standardization sample. In the present analysis, we assessed motor impairment in children with balance abilities, using two values, standardized and percentile scores. The physiotherapists in our unit regularly assess the balance and aiming subsections because these motor aspects are closely related to their field of competence. All of the scores of these clinical tests are reported and analyzed to evaluate the delay in motor development. In our current study we based our analysis on the score of MABC-2 Balance. All included subjects were successfully assessed using this test.

2.2.3. Postural recordings

Postural control was evaluated for each child, using Multitest-Equilibre, also called Balance Quest, by Framiral (www.framiral.fr). We assessed the PC on a stable and unstable support (respectively, S and U) in several conditions: eyes open (EO), where the children had to focus on a small light found at 250 cm in front of their eyes; eyes closed (EC), and the OKN condition was performed with the eyes open with optokinetic stimulation provided by a planetarium projected on a wall at a distance of 250 cm from the children with a mean rotation velocity of 15°/second. The entire procedure has already been extensively described elsewhere [22-23]. To obtain the vestibular score in each sensorial condition the classic postural parameter was used: surface of Centre of Pressure (CoP), which corresponds to an ellipse with 90% of CoP excursions and is therefore highly representative of the spatial variability of postural sway [24]. This parameter was coupled with the analysis from temporal postural parameter: the spectral power index in the postero-anterior direction. The smaller the spectral power index, the better the postural stability. This parameter is measured on three frequency bands (Low frequency band 0.05-0.5 Hz, Medium frequency band: 0.5-1.5 Hz, and high frequency band: greater than 1.5 Hz). It is more relevant to associate analysis of spatial postural parameter with temporal parameter since the analysis of spatial parameters alone is less discriminating and less precise and-reliable [25-27].

2.3. Statistical analysis

We performed the impact of saccular response on postural abilities recorded with Multitest-Equilibre platform as well as with clinical motor function (MABC-2). We performed an ANOVA, with post hoc LSD on the IBM SPSS Statistics 21 software. The criteria for grouping was saccular function: group 1 (normal bilateral), group 2 (normal unilateral) and group 3 (absent bilateral).

3. Results

3.1. Results are shown in **Table 1** for the group with normal bilateral saccular responses (Group 1), the group with unilateral saccular response (Group 2) and the group with absent bilaterally saccular response (Group 3). **Figure 2** shows the scores MABC-2 balance in children depending on saccular function. Balance score was found at 25.5 in Group 1, 16 in Group 2 and 14 in Group 3.MABC-2 balance score is significantly greater in children with normal bilateral responses in comparison to those with unilateral saccular response (p=0.02) and without saccular response (p=0.01).ANOVA showed a significant effect of saccular function $F_{(2,25)}$=5.09(p=0.01).

3.2. The surface of CoP (cm²) is shown in **Figure 3**, in OKN-U condition, in children depending on saccular function. The surface was 16.2 cm² in Group 1, 18.6 cm²in group 2 and 28.8 cm² in group3.Surface of CoP in OKN-U condition was significantly smaller in group 1 in comparison to group 3 (p=0.01). ANOVA showed a significant effect of saccular function on the surface of CoP, in OKN-U condition, $F_{(2,75)}$=3.75(p=0.02).

3.3. **Figure 4** shows the spectral power index (log), in OKN-U condition, in all ranges of frequency band (low, medium and high) depending on saccular function. For these frequencies' indexes were respectively 93, 85.7 and 72.5 in Group 1;104, 97 and 87 in

Group 2 and 129.6, 123 and 115 in Group 3. Spectral power index in OKN-U condition was significantly greater in group 3 in comparison to those in group 2 in low, medium and high frequency (all p=0.01) and group 1 (p=0.03 in low frequency and p=0.04 in medium and high frequency). ANOVA showed a significant effect of saccular function on spectral power index, in OKN-U condition, $F_{(2,75)}=3.96$; p=0.02; $F_{(2,75)}=3.48$; p=0.03 and $F_{(2,75)}=3.71$; p=0.02, respectively in low, medium and high frequency.

4. Discussion

The main findings from this study conducted on hearing impaired children, are the following: *(i) bilateral presence of cVEMPs was correlated with a good postural control on the Balance Quest Platform and higher scores of MCME -2 test; (ii) although not significant, unilateral absence of cVEMPs was correlated with lesser equilibrium scores whereas bilateral absence of cVEMPs predicted a very poor quality of the PC; and (iii) both Dynamic Posturography and MCBE-2 assessments appeared to be appropriate to discriminate children with sacculo-otolithic deficits;*

4.1. The first finding is prominent when we look at the results and at the diagrams shown in Figures 2 to 4. Both clinical motor tests using MABC-2 and postural sway recordings with Multitest-Equilibre including spatial and temporal parameters in this series, reported an impact of sacculo-collic deficiency on postural stability. De Kegel et al. also found a poorer performance in three subsections of MABC-2 in children with hearing impairment, compared to a group of age-matched typically developing children [10]. In their study, the authors conducted a vestibular assessment using cVEMPs and a rotary chair; they found that cVEMPs and rotary chair testing had predictive scores of the balance performance of hearing-impaired children.

Normal vestibular function enables gaze stabilization, sense of the verticality and of self-motion [28]. Consequently, a vestibular dysfunction would generate postural inability, poor visual dynamic acuity and spatial disorientation which are premises for a delayed or a poor posturomotor development especially in young children [5].

As previously emphasized, the otolith function would mature independently of the canal function and may be primarily linked to motor abilities while learning to walk [6, 29]. This hypothesis was supported in a later retrospective study of the same group [7]. Following these results, our study showed that children with normal bilateral saccular responses (Group 1) had a significantly higher score in MABC-2 and better dynamic posturography performances compared to children with no saccular response (Group 3);

4.2. Although non-significant, a tendency to a lower PC was also observed in the unilateral saccular response subjects (Group 2).

4.3. Recently, Oweyumi et al. compared children with bilateral vestibular dysfunction and normal, describing a quick and relevant screening to discriminate the subjects with poor PC [30]. Their retrospective study reported that the inability to perform standing on one foot with the eyes closed for more than 4 seconds was a very sensitive and discriminating test to support a suspicion of vestibular dysfunction. In this sensorial paradigm, somesthetic input is diminished and visual cue is unavailable. Consequently, vestibular input should be prioritized by the PC in order to preserve the upright standing position of the subject. This explains in the case of vestibular hypofunction why this sensory condition is so challenging and those subjects so unstable and failing to keep standing on one foot no more than 4 seconds. In the daily office practice, a clinical maneuver as above, especially when coupled with the MACB – 2 test allows to assess with accuracy the quality of the PC during childhood and therefore, to suspect a potential vestibular impairment.

Through our current results, we found that clinical motor tools MABC-2 and posturographic Balance Quest Platform assessment are both sensible to indicate a possible sacculo-otolithic dysfunction. Although it appears not to be sensible enough at the time of this study, temporal parameter with spectral power index from posturographic recording could be, in the future, more specific to indicate subjects with unilateral saccular deficit. To the best of our knowledge, this potential sensitivity has not been yet hypothesized and would allow improving the clinical evaluation in the future. Following previous reports, our study also confirms that children with hearing impairment frequently show various degrees of vestibular impairment although symptomatology may be poor [31].

The therapeutic approach in children with profound hearing loss is the cochlear implantation (CI) (uni- and/or bilateral) shortly after diagnosis. It is well established that CI surgery could damage the vestibular structures and namely the saccule [32]. In the light of our results, it appears that children candidates for a CI should categorically receive a vestibular assessment before surgery since nowadays this is not a difficult task for specialized teams after a short period of training. Consequently, due to non-negligible risks although soft surgery techniques are well quantified, surgeons should avoid performing bilateral CI in onetime surgery in young children whose walking is not yet acquired. Instead, this approach should be considered as previously advocated in infants with severe bilateral vestibular impairment [33]. Such decision should be therefore supported by both Audiological and Vestibular assessment.

In case of unilateral post CI vestibular impairment, vestibular rehabilitation therapy should be advocated to diminish the risk of delayed motor abilities which frequently occurs in these subjects. Special attention should be given in bilateral vestibular impaired children in order to improve their postural control strategies which are severely impacted.

From a practical point of view these findings strongly suggest that all very young CI candidates, especially those that cannot walk should systematically undergo vestibular assessment before surgery. Taking these precautions would diminish the risk of bilateral labyrinthine failure and its consequences such as vestibular ataxia and oscillopsia resulting in significant delays in posturo-motor milestones.

5. Conclusion

5.1. Bilateral absence of sacculo-collic function is pejorative for the postural control and the motor development in hearing impaired children.

5.2. Subjects with one available sacculo-collic function may show quasi normal or slightly poorer postural performance.

5.3. Surgeons should be aware about the negative consequences of the bilateral saccular deficit on the postural control, when considering bilateral CI at the same surgical especially in children who haven't yet started walking.

6. Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Informed consent was obtained from all individual participants included in the study.

7. Competing Interests

The co-authors declare that they have no competing interests.

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Legends

Figure 1:

Figure 2: Mean MABC-2 balance scores in children depending on saccular function: with normal bilateral saccular responses (Group1), with unilateral saccular response (Group 2) and bilateral absent saccular response (Group3).

Figure 3: Surface of CoP (cm^2), in OKN-U condition, in children depending on saccular function (Group1, Group 2 and Group 3).

Figure 4: Spectral power index (log), in OKN-U condition, in all bands frequency (low, medium and high) depending on saccular function in all groups (Group1, Group 2 and Group 3).

Table 1: Composition of the three groups and main results with mean scores: Standard Error (SE), Standard Deviation (SD).

Figure 1

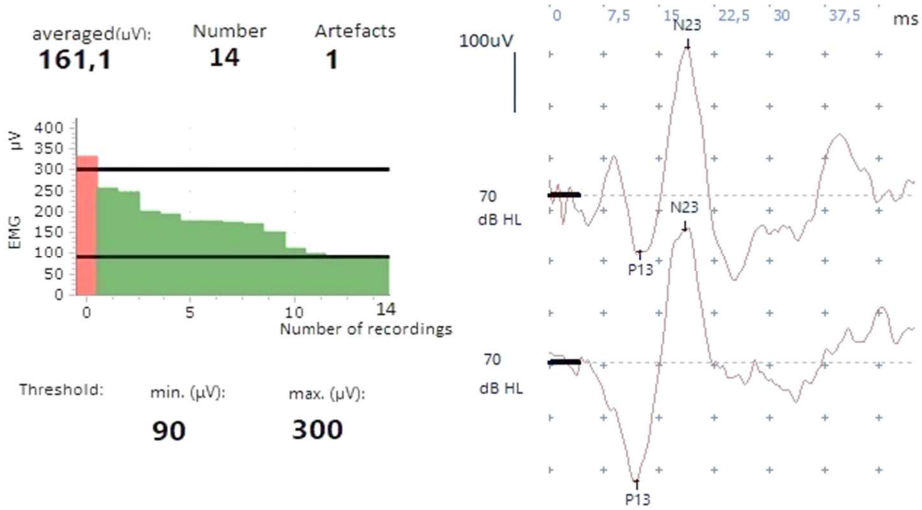


Figure 2

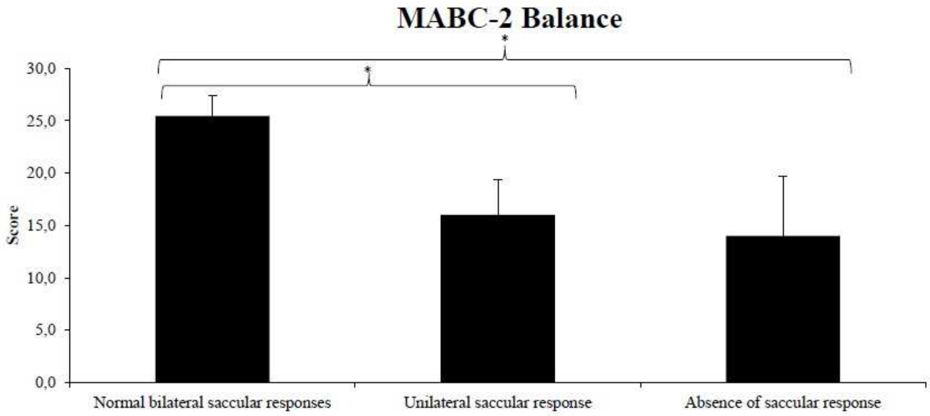


Figure 3

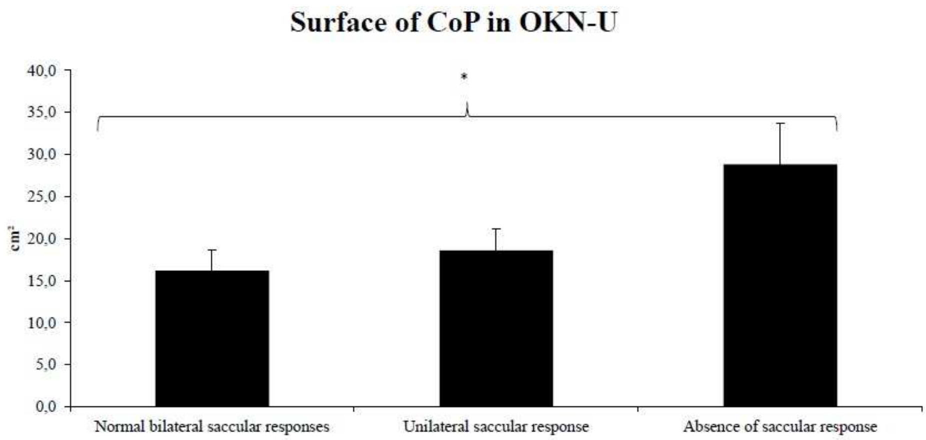
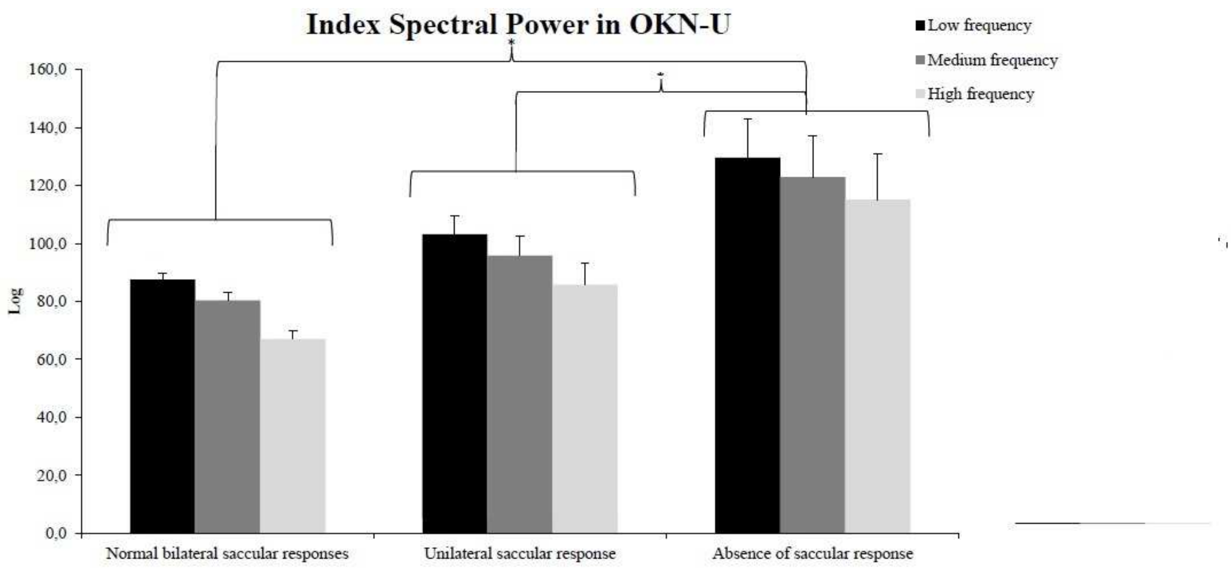


Figure 4



Legends

Figure 1: Normal biphasic characteristics of P13-N23 cVEMPs waves elicited by 500Hz tone burst bone stimulation at 70dB HL. EMG signals were monitored, amplified and bandpass filtered between 30 and 3000 Hz.

Figure 2: Mean MABC-2 balance scores in children depending on saccular function: with normal bilateral saccular responses (Group1), with unilateral saccular response (Group 2) and bilateral absent saccular response (Group3).

Figure 3: Surface of CoP (cm²), in OKN-U condition, in children depending on saccular function (Group1, Group 2 and Group 3).

Figure 4: Spectral power index (log), in OKN-U condition, in all bands frequency (low, medium and high) depending on saccular function in all groups (Group1, Group 2 and Group 3).

Table 1

							Index spectral power		
	Mean age	Sex ratio M-F	Amplitude cVEMPs OD	Amplitude cVEMPs OG	MABC-2 (balance)	Center of pressure (cm²)	LF	MF	HF
Group 1 n=41		26M- 15F							
mean	8.8		360	342.4	25.5	16.2	93	85.7	72.5
SD	3.3		163.25	156.2	7.7	16.2	9.5	11.2	11.43
SE	0.5		25.8	24.7	1.98	2.5	2.23	2.64	2.7
Group 2 n=18		12M- 6F							
mean	9.6		177	178.2	16	18.6	104	97	87
SD	3.1		201.9	195	8.4	10.8	40.8	44	48.3
SE	0.73		50.5	52	3.4	2.56	6.4	6.9	7.5
Group 3 n=17		10M- 7F							
mean	9.4		0	0	14	28.8	129.6	123	115
SD	3.8		0	0	11.4	20	54.4	59.4	65.1
SE	0.93		0	0	5.7	4.87	13.2	14.4	15.7

Table 1: Composition of the three groups and main results with mean scores: Standard Error (SE), Standard Deviation (SD).