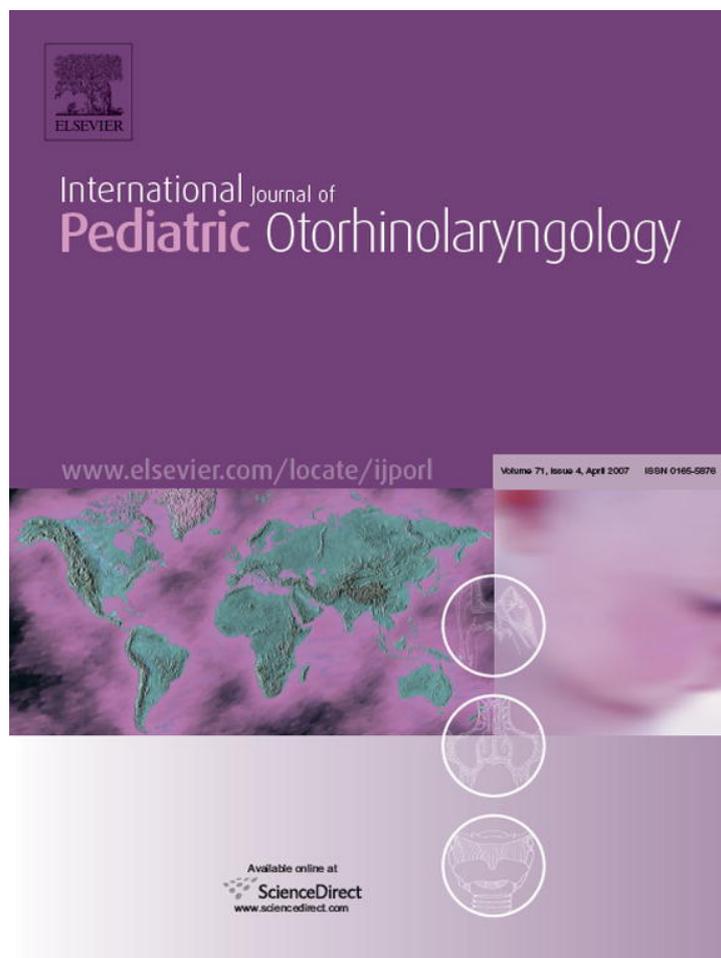


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Balance sensory organization in children with profound hearing loss and cochlear implants

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Summary

Objectives: (1) To determine the feasibility of the use of a modified postural control test under altered sensory conditions in children over 8 years of age, and (2) to assess how deaf children use sensory information for postural control when they have normal or abnormal vestibular responses, and if hearing input from a unilateral cochlear implant, changes their postural behavior.

Patients: We selected 36 children, 8 to 11 years of age, with congenital or early-acquired profound sensorineural hearing loss, 13 of them with unilateral cochlear implantation and 22 normal-hearing children.

Methods: The Postural Control (PC) test consists of a force platform with 2 stimulation paradigm conditions: (1) standing on the platform with opened eyes; (2) standing on foam placed on the force platform with closed eyes. Implanted children were tested with the implant turn on and turn off in this condition, in order to evaluate eventual change in the postural control parameters when they have hearing habilitation. The body center of pressure distribution area (COP) and the body sway velocity (SV) were the parameter to evaluate the postural control.

Results: Deaf children were classified into two groups according with the vestibular responses: group A ($n = 28$) Children with normal vestibular rotary responses; group B ($n = 8$) children with hypoactive responses. Children in group A had diagnoses of syndromic and non-syndromic hereditary deafness, and children in group B had inner ear malformations, post-meningitis deafness, and one child had non-syndromic hereditary deafness with hypoactive vestibular response. In condition 1, when vestibular, somatosensory and visual information were enabled, the COP and SV values did not show any statistically significant differences between groups A, B and control. In condition 2, when visual information was removed and the somatosensory

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input strongly modified by standing on the foam, group B showed significant higher COP and SV values than groups A and control ($p < 0.05$). In addition, the scalograms by wavelets of children in group B had higher amplitudes increasing the sway frequencies contents up to 3 Hz, not allowing them to maintain the up right stance in similar stimulation than in condition. Implanted children of the group A and B with the implant turn on, in the condition 2, did not show any significant difference in the SV, comparing when they had the implanted turn off. Group A $p = 0.395$ and group B $p = 0.465$ (Wilcoxon ranked test).

Conclusion: These findings allow us to confirm that this postural test can be performed in children over 8 years old. Also our results suggest that deaf children with associated hypoactive vestibular responses included in our study, despite the etiology of the deafness, primarily use visual and somatosensory information to maintain their postural control. Hearing habilitation with a unilateral cochlear implant has no effect on the observed sensory organization strategy.

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1. Introduction

The mature vestibular system is responsible for stabilizing the position of the eyes, head and body in space, and helps to maintain an upright stance. It is composed of two parts, each with different roles: (1) the vestibular–ocular system, responsible for visual stabilization; and (2) the vestibular–spinal system, which maintains the orientation of the body in space and contributes to the postural tone necessary for the acquisition of motor developmental milestones. The development and maintenance of postural stability is a multisystem process that does not depend solely on vestibular input. Maturation changes in other sensory systems (primarily visual and proprioceptive), central nervous system (CNS) processing, and coordination of motor output are responsible for the changes in postural skills observed through adolescence. From the sensory systems perspective, young children are dependent on the visual system to maintain balance; as they grow older, they progressively begin to use somatosensory and vestibular information until reaching full maturity by the age of ten. Similarly, the coordination of the motor response and the gait pattern reach adult-like maturity by seven to ten years of age [1–3].

Auditory deprivation from birth brings about functional plastic changes in the CNS. One of these changes is the activation of the “meaning brain areas” by different sensory sources as illustrated by the role of visual input in lip reading and sign language communication [4]. Significant plastic changes also occur in deaf patients who receive cochlear implants, as demonstrated by the auditory adaptation that occurs due to a modified peripheral frequency map [5] and by the activation of the brain areas necessary for auditory processing. Imaging technologies [6] and psychophysical testing [7,8]

have contributed to the understanding of the role of these plastic properties of the CNS in aiding communication in sensory impaired individuals.

Children with deafness are at risk of vestibular dysfunction because in some forms of inner ear deafness the damage extends to the vestibular receptor as well. There are reports of peripheral vestibular dysfunction and delayed postural control in some types of congenital or early-acquired deafness such as in inner ear malformations, meningitis, viral labyrinthitis, and some forms of hereditary deafness. Children with bilateral vestibular loss since birth or early life present with delayed gross motor development. These children stand and walk later than healthy children. However, the postural disturbances that result from isolated peripheral dysfunction are usually corrected by the time these children grow to be teenagers [9]. The postural disturbances are corrected through a process of compensation whereby input from proprioceptive, visual, and other sensory systems substitute for the absent peripheral vestibular input. The well known fact that postural recovery after vestibular lesions in adults occurs despite no regeneration or recovery of the vestibular apparatus indicates that changes in the CNS are likely responsible for the processing of substitutive sensory input that leads to this clinical recovery. As it is the case with central auditory plasticity, peripheral vestibular loss may bring about plastic changes in the CNS that are responsible for the adequate processing of substitutive sensory information necessary for the acquisition of postural skills in deaf children.

Although there are numerous reports of vestibular and balance dysfunction in hearing-impaired children found in the literature, most studies fail to control for type, degree and etiology of the hearing loss, as well as for other confounding variables [10]. The presence and severity of the peripheral vestibular dysfunction

seems to be associated with the etiology and severity of the sensorineural hearing loss; thus, vestibular dysfunction may be more prevalent among cochlear implant candidates than among children with lesser degrees of hearing loss. Furthermore, some forms of syndromic deafness have concomitant impairments in other sensory systems or in the CNS. Controlling for confounding variables is needed to adequately assess the contribution and integration of different sensory systems in postural control, particularly in the hearing-impaired child, where it is plausible that a reorganization of the sensory integrative properties of the CNS has taken place as an adaptation response to the sensory impairment.

This study was designed to examine how deaf children use sensory information for postural control, when they have normal or abnormal vestibular responses. We used a quantitative test of postural sway under conditions of missing or conflicting sensory information adapted from the sensory organization posturography test [11]. We wanted to assess if this test was sensitive to changes in sensory

organization strategies previously reported in children with various etiologies and degrees of hearing loss, and whether habilitation of hearing via a cochlear implant affected these changes.

2. Subjects and methods

This is a cross-sectional study of deaf and normal-hearing children eight to eleven years of age. We enrolled 36 children with congenital or early-acquired profound sensorineural hearing loss, 13 of them with unilateral cochlear implantation and 22 normal-hearing children. Demographics and clinical data are shown in Tables 1 and 2. We excluded children with associated cognitive or motor disabilities and children younger than 8 years of age because the Postural Control test used in this study has not been normalized for this age group. Neurological and visual acuity were normal in all the population studied. The 13 implanted children were ascertained at the Cochlear Implant Division of the

Table 1 Group A description

Age	Gender	Etiology	Condition 1		Condition 2	
			COP	SV	COP	SV
11	M	Conexin	3.12	1.75	3.73	5.02
11	F	Unknown	2.81	2.78	16.71	5.23
9	F	Unknown	3.9	1.85	35.31	5.97
10	F	Unknown	2.8	1.42	29.03	5.97
12	F	Conexin	2.87	1.71	12.19	2.27
10	M	Conexin	2.94	2.63	22.62	3.9
8	M	Unknown	2.4	1.78	11.93	3.41
9	F	Conexin	1.65	1.12	18.73	4.56
9	F	Conexin	1.82	1.32	19.67	4.87
11	M	Unknown	1.67	1.78	15.3	3.65
13	M	Unknown	1.43	1.22	18.76	4.12
9	M	Conexin	1.73	1.45	19.67	4.87
8	M	Unknown	1.58	1.74	15.3	3.65
10	F	Conexin	2.34	1.98	18.76	4.12
10	F	Conexin	3.2	2.19	24.83	4.56
12	M	Unknown	1.74	1.21	25.88	4.15
9	M	Unknown	1.43	1.18	14.72	3.12
13	F	Unknown	1.73	1.65	12.65	3.05
10	F	Unknown	2.38	2.23	25.12	4.54
10	M	Conexin	2.38	1.67	21.56	4.22
9	F	Conexin	1.46	1.74	29.52	4.56
12	F	Unknown	2.12	1.53	18.15	3.78
12	F	Unknown	1.45	1.14	17.46	3.95
8	M	Unknown	2.81	2.34	18.41	4.03
10	F	Unknown	1.96	1.64	20.16	3.53
11	F	Wallenberg	1.85	1.32	18.16	3.74
9	M	Wallenberg	1.87	1.78	19.09	4.01
11	M	Unknown	1.95	1.59	25.39	2.16

This group included profoundly deaf children with normal vestibular responses. Ethnic group: white. Body center of pressure distribution area (COP cm²) and body sway velocity (SV cm/s) values in conditions 1 and 2.

Table 2 Group B description

Age	Gender	Etiology	Condition 1		Condition 2	
			COP	SV	COP	SV
11	M	Posmeningitis	0.84	0.72	111.12	5.01
9	M	Posmeningitis	1.23	1.02	129.12	10.72
9	F	Bilateral inner ear aplasia	2.4	1.33	105.01	7.27
10	M	Posmeningitis	2.34	2.44	66.9	7.27
8	M	Unknown	0.91	1.08	162.4	13.38
11	F	Posmeningitis	2.34	1.79	167.15	9.57
11	M	Bilateral Mondri	1.79	1.54	142.24	11.9
10	M	Bilateral Mondri	1.64	1.23	97.34	6.2

In this group are deaf children with associated hypoactive vestibular responses. They are mainly children with inner ear malformations and post-meningitis deafness, is also included a child with non-syndromic deafness who showed hypoactive vestibular responses. Ethnic group: white. Body center of pressure distribution area (COP cm²) and body sway velocity (SV cm/s) values in conditions 1 and 2.

British Hospital of Montevideo and School Of Medicine between January 2004 and December 2005 and in all of them the cochlear implant was activated a month after surgery. The study subjects underwent an evaluation which included a detailed medical and family history with emphasis on identifying manifestations of acquired or syndromic deafness, neurotological physical examination, age-specific audiological testing, and vestibular evaluation. Vestibular evaluation was performed in the implanted children at least six months after implant activation. Passive whole body testing [12] with ENG record was performed in order to determine the vestibular function, using low frequencies <0.05 Hz, according to Hess et al. [13] normal values determinations. Deaf children underwent high-resolution 1.5 mm-cut computed tomography of the temporal bone (CT) and peripheral blood DNA

testing for *GJB2* gene mutations. Institutional review board approval for the genetic studies and appropriate informed consents were obtained.

Screening of *GJB2* gene mutations: Nuclear DNA samples from peripheral leukocytes were screened for the 35delG mutation by an allele-specific PCR test. In heterozygous and those with negative 35delG mutations, the coding region of the *GJB2* gene was screened by direct sequencing approach [14].

Postural control was assessed with the Postural Control test (PC). This test is based on the principles of the Test of Equilibrium Under Sensory Altered Conditions [15] (Fig. 1). The stimulation paradigm was as follows: Condition 1: The patient is in standing position on a force platform with opened eyes. Condition 2: The patient is in standing position on a 20 cm-thick foam placed on the force platform with

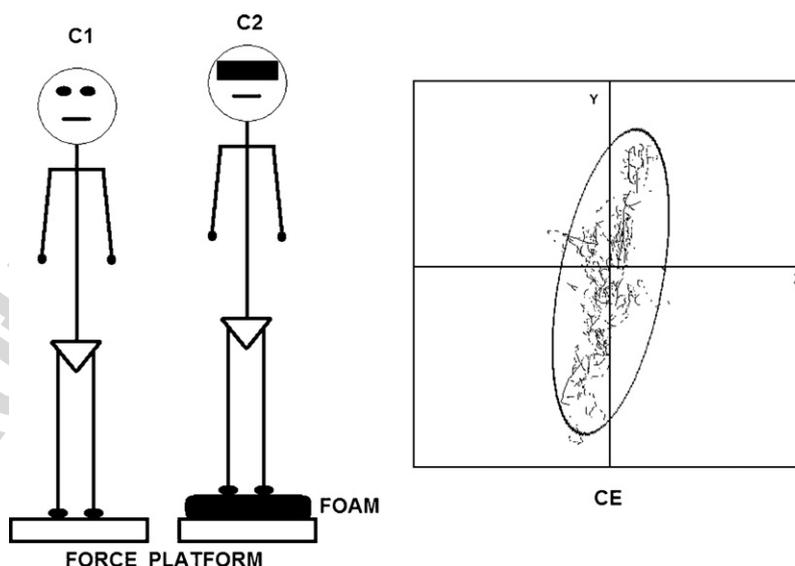


Fig. 1 Schematic representation of the stimulation paradigm conditions in which were recorded the postural responses. C1 – condition 1, visual, vestibular and somatosensory information. C2 – condition 2, absence of visual input, modified somatosensory, and normal vestibular information. CE – confidential ellipse of the COP area.

covered and closed eyes. In this condition the cochlear implanted children had their implant turned off and turn on exposed to environmental noise.

Condition 1 explores postural control while enabling input from visual, vestibular and somatosensory end-organs. In condition 2, visual information is eliminated and, by using the thick foam, somatosensory input is altered therefore the subject has to rely primarily on vestibular information during the PC test. In implanted children they were evaluated in presence or absence of additional sourced of sensory information when the cochlear implant is turn on or turn off, respectively.

Posture control strategies were assessed using a force platform with online recording of the body center of pressure measuring two relevant parameters: sway velocity (SV) and the 95% confidential ellipse of the center of pressure (COP) distribution area.

SV. A 80 s trial was recorded producing 2 discrete signals of $n = 4000$ samples (sampling frequency $f_s = 50$ Hz): COPx and COPy. Then, for each recording, the average speed of COP along its path ($\langle v \rangle$) was calculated at $t = 80$ s ($n = 4000$) using:

$$\langle v \rangle = \frac{f_s}{N} \sum_{i=2}^N [(COPx_i - COPx_{i-1})^2 + (COPy_i - COPy_{i-1})^2]$$

2.1. Confidential ellipse (CE)

The CE of the bivariate distribution $(COPx_i, COPy_i)$, $1 \leq i \leq N$, is the ellipse where 95% of the COP samples are predicted to be enclosed. The area of the 95% confidential ellipse is calculated as follows:

$$\text{Area} = 2\pi F_{0.05[2, N-2]} \sqrt{\sigma_x^2 \sigma_y^2 - \sigma_{xy}^2}$$

where $F_{0.05[2, N-2]}$ is the F statistic at the 95% confidence level with N data points, σ_x^2 and σ_y^2 are the variances of the ML and AP coordinates, respectively and σ_{xy} is the covariance. For a large sample size ($N > 120$), $F_{0.05[2, N-2]}$ is 3.00. This is the case here ($N = 4000$).

2.2. Time-frequency analysis (scalogram)

In order to evaluate the fundamental oscillatory frequency, its amplitude and temporal behavior of the responses, a time-frequency analysis of COP in both directions (COPx and COPy) was performed by computing its scalogram. As the Fourier Transform is not adapted to the analysis of non-stationary signal such as the COP signal, its time-frequency representation must be considered.

Because of its resolutions properties, a widely-used time-frequency energy density is the scalogram. The scalogram of the signal $x(u)$ is the energetic version of the wavelets transform (WT), defined as the square magnitude of WT:

$$\text{SCAL}_x(t, f) = \left| \int_0^{+\infty} x(u) \sqrt{\frac{f}{f_0}} \psi \times \left(\frac{f}{f_0} (u - t) \right) du \right|^2$$

The mother wavelet that was chosen was the Morlet wavelet:

$$\psi(u) = e^{-u^2/2} e^{j2\pi f_0 u}$$

This wavelet is the one with the best time-frequency localization in the sense specified by the Heisenberg-Gabor uncertainty principle [16].

Statistical Analysis: The Kruskal–Wallis test was used for comparing mean values of CE and SV among groups.

Wilcoxon rank sum test was used to compare postural behavior in implant children, when the implant is “turn on” and “turn off”, with a significance α level of 0.05. SV was used for statistical analysis, because it shows more sensibility for postural control [17].

3. Results

Deaf children were further classified into two groups according to the vestibular responses: group A ($n = 28$): children with normal vestibular rotary responses. Group B ($n = 8$): children with hypoactive responses. Children in group A had diagnoses of syndromic and non-syndromic hereditary deafness, and children in group B had inner ear malformations, post-meningitis deafness, and one child had non-syndromic hereditary deafness. The child with a non-syndromic hereditary deafness was included in group B because he had hypoactive vestibular response.

3.1. PC behavior in condition 1 and 2 of the stimulation paradigm

The COP and SV values were higher in condition 2 than in condition 1 both in the deaf children population, groups A and B (Tables 1 and 2), and in the normal control group. Assuming that in condition 2 the visual input is lost as information source, and somatosensory information is strongly modified, vestibular ends organs have the main role in maintenance of the PC.

Group A: No significant differences to control children were found in the PC behavior in conditions 1 and 2 (Fig. 2). The etiology of the deafness in this

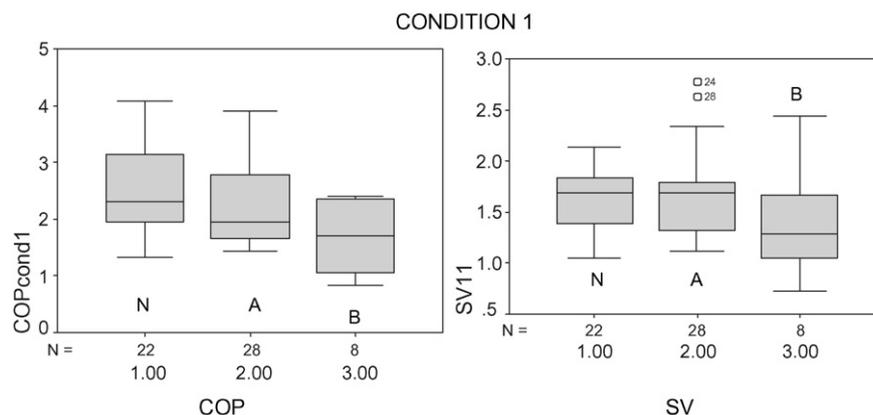


Fig. 2 Box-plot of the median values of the 3 groups in condition 1: (N) normal control group; (A) deaf children with normal vestibular rotatory responses; and (B) deaf children with hypoactive vestibular rotatory responses. The top and bottom of the box represent the first and third quartile of each sample, though the 50% of the sample values are in the “box”. The top and bottom bars represent the maximum values for each group. The small squares are considered outliers values of the sample. Not significant differences in the mean COP (left) and SV (right) values. Kruskal–Wallis.

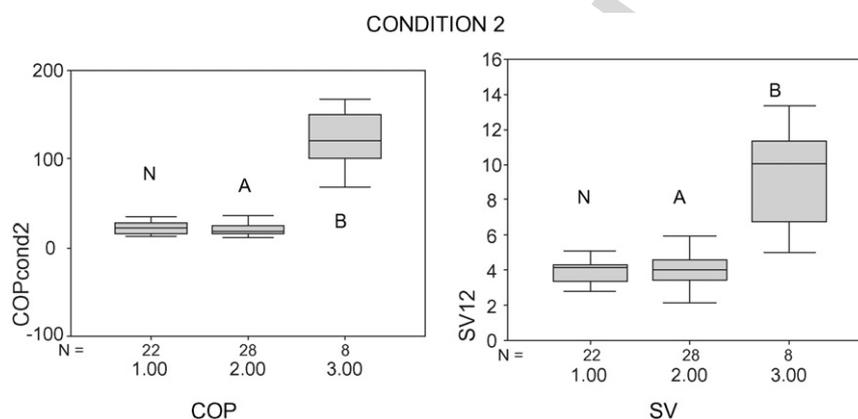


Fig. 3 Box-plot (see text of Fig. 2) of COP (left) and SV (right) values in condition 2: comparison between (N) normal control group; (A) deaf children with rotatory responses and (B) deaf children with hypoactive vestibular rotatory responses. Group B had significant higher values – Kruskal–Wallis – in the COP and SV values.

group was congenital non-syndromic deafness in 16 patients, Connexin 26 deafness in 10 patients, and Waardenburg syndrome in 2 patients (Table 1).

Group B: These children had difficulty in maintaining the standing position in the paradigm condition 2, presumably due to their vestibular hypofunction. This group included 4 patients with post-meningitis deafness, 3 patients with inner ear malformation, and 1 patient with congenital deafness of unknown etiology (Table 2). The COP and SV values for condition 2 of this group were significantly higher (Kruskal–Wallis test) than the values of both the control group and group A (Fig. 3). The scalogram by wavelets in condition 2 showed that children of group B had higher sway frequencies contents and amplitudes than group A (Fig. 4).

Children in group B could not maintain the standing position during the time of recording (80 s) when the visual information was removed and the soma-

tosensory input was modified, suggesting that the vestibular end organs are not providing enough sensory information to maintain stance during PC recordings in condition 2.

The 13 implanted children of the group A and B did not show any significant difference in the SV, with the implant “turn on” and “turn off” (Wilcoxon Ranked test) in the condition 2 of the paradigm stimulation (Table 3).

4. Discussion

Children with congenital and early-onset deafness are at risk of concomitant peripheral vestibular loss. Despite this vestibular loss, most deaf children eventually reach an adequate postural and gait control through the use of substitutive strategies. The plastic changes that occur as a consequence of

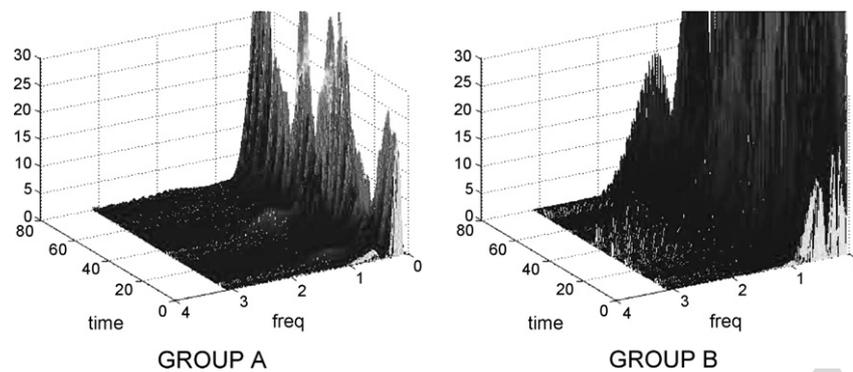


Fig. 4 Scalogram by wavelets. Typical case of group A (left) and group B (right) in condition 2. Group B shows responses with higher amplitudes and sway frequencies contents up to 3 Hz precluding the maintenance of the upright stance.

early deafness, such as the activation of the “meaning brain areas” by visual cues and neural adaptation to recognize electronic signals from a cochlear implant, illustrate that the CNS also adapts to auditory deprivation by the use of substitutive strategies. This study shows that older children with early deafness and vestibular loss (group B) depend on visual and somatosensory information to maintain their postural control. The test of PC used in these experiments showed to be sensitive to the identification of substitutive sensory strategies for postural control. When deaf children with vestibular dysfunction (group B) were exposed to the stimulation paradigm, condition 2, in which the visual stimuli is removed and the somatosensory input is strongly modified (therefore the vestibular input is the main source of balance information) they showed abnormal COP and SV values precluding the maintenance of the upright stance.

These children, however, do not show difficulty with tasks of daily living such as walking, running,

and participating in sports. These findings suggest a sensory substitution process whereby visual and somatosensory information becomes essential for postural control. It is remarkable that the visual and somatosensory substitution allowed normal postural skills in this group of deaf children (group B); when visual and somatosensory input were enabled, as in condition 1, these children showed COP and SV values similar to those of deaf children with normal vestibular function as well as those of healthy children.

Vestibular dysfunction is found in approximately 20–70% of children with hearing loss of different causes [18]. Furthermore, the prevalence of vestibular tests abnormalities is higher in profound versus severe SNHL, [19,20] in post-natally acquired cases (meningitis, labyrinthitis), [18–20] in some forms of syndromic deafness (Usher, Waardenburg, and Pendred syndromes), [18,21,22] and in children with labyrinthine dysplasia [23]. In contrast, recessive inherited deafness is usually associated with normal vestibular function [18].

Rine et al. [24] studied gross motor development and vestibular function in a group of 24- to 83-month-old children with SNHL. They measured nystagmus duration after rotatory stimulation to assess vestibular function, and performed repeat motor and vestibular testing in a sub-group of children. These authors concluded that children with SNHL and concurrent vestibular hypofunction are likely to show a progressive delay in gross motor development, and that normal post-rotatory nystagmus testing has an excellent sensitivity for the identification of children most likely to present with normal motor development. They recommend testing for vestibular hypofunction in children with SNHL as early as possible in the preschool years, so treatment can be instituted in a timely fashion.

To adequately determine if the balance and motor delay observed in children with SNHL is due to vestibular dysfunction, both arms of the

Table 3 Implanted children of groups A (left) and B (right)

Sway velocity			
Group A		Group B	
Off	On	Off	On
1.33	1.36	10.52	10.54
2.44	2.41	13.38	13.5
1.08	1.12	3.41	3.41
1.78	1.73	3.65	3.62
1.78	1.82	4.15	4.17
1.72	1.69		
1.23	1.26		
1.79	1.86		

SV (cm/s) values in the implanted children exposed to condition 2. In both groups SV (raw data of each patient), values with implant off (left column) and on (right column). There is not significant statistical difference with the implant on or off, in both groups. (Wilcoxon Ranked test). Group A $p = 0.395$, group B $p = 0.465$.

vestibular system (VOR and vestibular–spinal function) should be evaluated. Thus, tests that quantify postural sway under different sensory conditions are indicated, particularly since the bedside clinical test of postural control, the Romberg test, is not sensitive enough to diagnose most cases of vestibular dysfunction [25]. The sensory organization test of dynamic posturography serves to identify the area of deficit and aids in programming the strategy for rehabilitation [10,25].

Some authors have advocated the determination of vestibular dysfunction in congenitally deaf children for the design of a deficit-specific rehabilitation program [21,25,26]. Children with bilateral vestibular loss are taught to use substitutive strategies (sensory and motor); conversely, vestibular habituation exercises are avoided since they are clearly ineffective in the subject without vestibular function. In terms of counseling and prevention, children with bilateral vestibular dysfunction need to recognize dangerous situations in which both the visual and proprioceptive systems are rendered unreliable, such as swimming in the dark.

Additional recommendations for the vestibular-deficient child include the correction of concomitant deficits in participating systems (e.g., vision correction), and the optimization of sensory input: avoidance of thick soles for the more effective use of pressure-receptors of the feet, incremental experience with ambulation which stimulates sensory organization.

Although further research will be necessary, the non significant differences in the SV in the condition 2 when the implant is “turn on” and “turn off”, suggest that the auditory information from a unilateral cochlear implant is not contributory for postural behavior.

In conclusion, the test of postural control evaluated in this study is sensitive to identify substitutive sensory strategies for postural control in deaf children with and without labyrinthine hypofunction; children with hypoactive vestibular responses primarily use visual and somatosensory information to maintain their postural stability. Hearing habilitation with a unilateral cochlear implant has no effect on the observed sensory organization strategy.

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