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Saccular function evolution related to cochlear implantation in hearing impaired children

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Abstract

Vestibular sensorial input is essential for psychomotor development of the very small children. In consequence, possible vestibular impairment induced by cochlear implantation in deaf children could affect the balance and walking learning process. Some of cochlear implanted children can present congenital vestibular deficit. The anatomical and embryological relation between auditory and vestibular system explains why congenital neurosensorial hearing loss may associate vestibular impairment. The cochlear implant surgery presents a vestibular lesion risk. Bilateral vestibulopathy, as it appears in early childhood, has a poor prognosis for the psychomotor and cognitive development. Even probably rare, bilateral vestibulopathy induced by simultaneous bilateral cochlear implantation can delay the acquisition of motor skills. This pathology can be avoided by an appropriate surgical indication related to the vestibular preoperative status. This study reports the vestibular saccular functional modifications after the cochlear implantation in children. The cervical vestibular evoked myogenic potentials (cVEMPs) were performed in children before and after the cochlear implantation. Since previous studies report different vestibular impairment related to the portelectrode insertion approach, another objective of our study was to assess the saccular postoperative status depending of the insertion by cochleostomy (CO) or through the round window (RW). We performed cVEMPs for 80 patients (135 cochlear implanted ears) before and after cochlear implantation. We have detected preoperative saccular areflexia in 33 (24.4%) ears. In the group of 102 (75.6%) ears with preoperative normal saccular function, 72 (70.6%) ears preserved the cVEMP response after the surgery, while in 30 (29.4%) ears the cVEMP response was lost. Reporting our findings to the portelectrode insertion method, we found normal saccular function in 73.3% of the cochlear implanted ears by RW surgical approach and in 68.42% ears by CO approach. These results suggest that the RW portelectrode insertion is the recommended strategy in order to avoid the saccular vestibular impairment.

Keywords: saccular vestibular function, vestibulopathy, cochlear implant, cochleostomy, round window.

Introduction

Cochlear implant (CI) is the gold standard treatment for profound and severe deafness that cannot be adequately corrected by hearing aids. As a result of the reported performances in the auditory rehabilitation, the indication of the CI has progressively expanded, including more and more typologies of patients.

This type of treatment represents the only one solution especially for the children with bilateral congenital or progressive neurosensorial hearing loss (NSHL). The bilateral implantation in children is more and more indicated in children than unilateral implantation due to the important advantages increasing the life quality: giving the possibility of the sound localization, improving the hearing in noise and the speech understanding, ensuring the auditory perception in case of one implant's failure and favoring the balance system's development.

One of the reported complications of CI surgery is the vestibular damage, which can be transient or permanent. Previous researches reported that the surgical insertion of the portelectrode induces a various vestibular damages in 50% to 85% of the implanted cases [1]. A recent review confirms that the postoperative vestibular lesion's rate is highly variable, between 18% and 85% of the implanted children. The mechanism could be represented by the direct trauma induced by the insertion of the portelectrode or by other pathological processes. Different morpho-pathological studies mention local infections and ischemic or hemorrhagic processes as determinant factors for the endothelial vestibular lesions. The presence of the port-electrode in the internal ear space may produce autoimmune reactions or endolymphatic hydrops. At the end of the surgical intervention, the sealing of the cochleostomy (CO) or of the round window (RW) is important to avoid a perilymphatic fistula, witch could itself induce a vestibular syndrome. It is also reported in the literature the vertigo induced by electrical stimulation through the portelectrode [2–4].

An important factor that may influence the preservation

of both cochlear and vestibular neurosensory epithelium is the surgical approach. There are two ways to rich the intracochlear space: through the RW and by CO. The literature presents different results regarding the preservation of the vestibular function using these two methods.

Some of the children with profound congenital NSHL may present as well congenital vestibular impairment, due to the anatomical and embryological relation of the different parts of the inner ear. The prevalence of the vestibular dysfunction in children with NSHL ranges from 20% to 85% [5–7]. This variability is related to the different associated conditions and pathologies. For example, in the group of genetic non-syndromic hearing loss, the mutations of deafness, autosomal recessive (*DFNB) 3*/*POUF 4* and *DFNB 4* associate different degrees of vestibular deficit, *DFNB 1* rarely affects vestibular function, while *DFNB 9* has exclusively auditory impairment. Syndromic NSHL is often associated with unilateral or bilateral, partial or complete vestibular deficiency. The most common syndromes with audio-vestibular impairment are: Usher, Jervell and Lange Nielsen, CHARGE, Waardenburg, Pendred, Goldenhar, DiGeorge. Isolated malformations of the internal ear like the lack of cochlear partition (Mondini syndrome), the single vestibular cavity (Michel syndrome), the enlarged vestibular aqueduct expresses themselves with cochleo-vestibular damages [8]. Congenital citomegalovirus infection is a risk factor for hearing loss and for vestibular lesions also. Ototoxicity of pharmaceutical substances (Gentamicin) can leads to hearing loss and bilateral vestibulopathy [9–12].

The occurrence of the vestibular deficiency in childhood, especially the bilateral one, leads to a chronic instability that will affect the child's motor and cognitive development. The prognosis is more severe if the vestibular deficit is present before the age of one year, because the child has not yet developed his walk and balance abilities [13, 14]. In these cases can appear axial hypotonia, problems of spatial and body representation. Cognitive loss could be generated by errors in building the self-image through relationship with the others and the space [15–17].

Aim

Taking in consideration that bilateral vestibular deficit represents a major negative impact for balance and global development, the rehabilitation of hearing loss by cochlear implantation should purpose to avoid to induce, if possible, any vestibular injury. To rich this objective, the vestibular status has to be assessed preoperatively. In practice, the cervical vestibular evoked myogenic potential (cVEMP) is the most used vestibular test in children, possible from 2–3 months of age. It is an objective, fast and non-invasive test, but some difficulties can appear when the child does not cooperate or in case of certain pathologies like neuropathies or muscular dystrophies.

Patients, Materials and Methods

To develop this prospective study, we enrolled 80 children (41 boys and 39 girls). Fifty-eight (72.5%) came from urban areas and 22 (27.5%) from rural areas. The mean age at the implantation moment was 4.35 years. The children were implanted for profound or severe hearing loss. Thirty percent had a monolateral implantation, 50% had a bilateral sequential implantation and 20% a bilateral simultaneous one.

The surgery was performed by the same surgeon for all cases. The cochlear devices were provided by Cochlear, MED-EL and Oticon. The surgical approach followed the common steps: retroauricular incision, mastoidectomy, posterior tympanotomy and portelectrode insertion. In order to respect the inner ear functional structures, the portelectrode insertion was done by atraumatic CO or by RW approach, depending also of the local anatomy.

After the surgery, a modified Stenvers radiography (oblique radiographic projection specific for the temporal bone) was done to verify the right position of the receiver-stimulator and intracochlear electrodes. The preoperative test protocol (T0) included the vestibular assessment of the saccular function by cVEMP. In order to avoid influencing the vestibular test through an external and/or a middle ear pathology, we performed a prior clinical examination of the ear by otomicroscopy and impedancemetry. The cVEMP was repeated postoperatively for each implanted ear in order to evaluate the preservation of saccular otolithic function. The test was scheduled at least three months after the surgery (T1) to avoid any transient vestibular deficit. For the cVEMP recordings, the child was placed in a sitting position in the arms of a parent, with the head turned toward the opposite side of the tested ear, looking to an interesting target. The sound stimulation was presented by air conduction through insert ear phones using tone burst with the following parameters: frequency of 500 Hz at the intensity of 100 decibels normal hearing level (dB nHL), duration of the stimulus of 2 ms, the rate of the stimulation 5.1 stimuli/s, the number of stimuli between 150 and 200 per run. We used the Eclipse evoked potentials device from Interacoustics, Denmark. Ipsilateral myogenic evoked potentials were recorded by placing the active electrode on the inferior third of the sternocleidomastoidian muscle (SCM), the ground electrode on the forehead, and the inverting electrodes on the retro-auricular areas. The patient must be kept in the mentioned position during the sound stimulation in order to maintain SCM's contraction. For cochlear implanted patients, the sound processor was removed from the head before performing the cVEMP test.

Until now, there is no consensus for normative parameters for the interpretation of cVEMPs in children. On the other hand, recording VEMPs in small children may present particular difficulties like the insufficient or inconstant contraction of SCM or the lack of compliance. For these reasons, we decided to report only the presence or the absence of the positive–negative (P1–N1) complex without any evaluation of the variation of its interpeak amplitude or wave's latencies.

Results

We analyzed first the preoperative saccular status in all 135 ears (defined as moment T0). In 75.6% of measurements, we obtained a cVEMP response, while in 24.4% of ears the saccular response was not present.

After the CI surgery (defined as moment T1), all implanted ears were retested in order to verify the conservation of the preoperative saccular status or to observe a

potential damage of the vestibular function. In 53.3% of the implanted ears, the cervical vestibular myogenic potential was present, while 46.7% of implanted ears do not show any saccular response (Figure 1). In order to highlight the real variation of the saccular function related to cochlear surgery, we should consider the saccular function variation only in the ears that in the preoperative moment had a present response for cVEMP. In consequence, in the group of 102 implanted ears with preoperative present cVEMPs, 70.6% preserved the saccular function after implantation, meanwhile 29.4% lost the vestibular potential. This variation may suggest the risk of injury that the surgical maneuvers for cochlear implantation and especially the intracochlear portelectrode insertion may have on the saccular neuroepithelium (Figure 2).

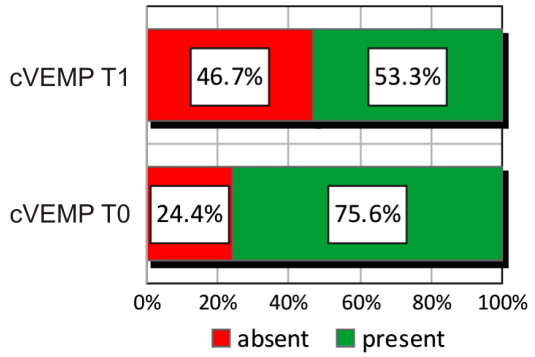

Figure 1 – ***Pre- and postoperative status of the saccular function in all implanted ears (n=135). cVEMP: Cervical vestibular evoked myogenic potential.***

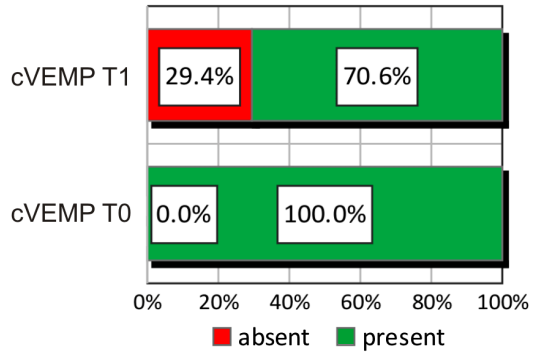

Figure 2 – ***Postoperative saccular function status (T1) in the group with preoperative present cVEMPs (T0) (n=102). cVEMP: Cervical vestibular evoked myogenic potential.***

The following chart (Figure 3) presents the variation of the vestibular saccular function in the group of 102 ears (who had saccular response preoperative) considering the surgical approach for portelectrode insertion: CO *versus* RW. In the CO group, considering the ears with present preoperative saccular response (57 ears), we found that 68.42% preserved this response, while 31.58% lost the saccular potential. In the RW approach group, selected by the same principle (ears that had a preoperative normal cVEMP), 73.33% maintained the physiological saccular function and 26.67% present saccular areflexia.

Being aware of the fact that the bilateral vestibulopathy can cause an unfavorable prognosis for the cognitive and the neuromotor development of the child, especially if it occurs concurrently on both ears, we analyzed in our group of bilateral cochlear implanted children how many among them present a bilateral saccular loss after the surgery in the respect of the time interval between the implantations. We defined two subgroups including children with at least one functional saccula at T0 who were bilaterally implanted, but differ by the surgery time: sequential bilateral surgery *versus* simultaneous bilateral surgery. In the first group that had a bilateral sequential cochlear implantation, there are eight (25%) patients that have a bilateral saccular lost after the surgery. In the second group that had a bilateral simultaneous implantation, two (18.18%) patients present bilateral cVEMP loss (Figure 4).

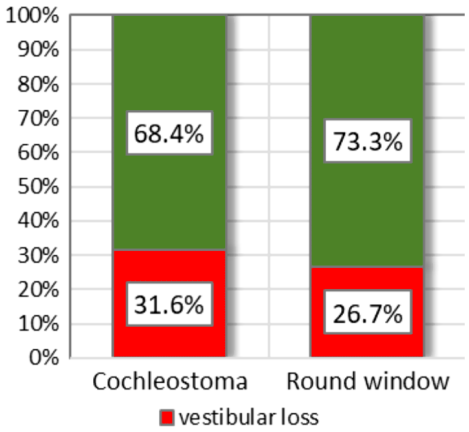

Figure 3 – ***Postoperative saccular status (T1) in the group with preoperative present cVEMPs related to the surgical approach (n=102). cVEMP: Cervical vestibular evoked myogenic potential.***

Discussions

The saccula is the closest vestibular structure to the cochlea and have a major risk of lesion, as the anatomo-pathological studies have been shown [18].

Although the CI surgery may induce different and permanent histopathological changes of the inner ear elements, there is no evidence of the vestibular nerve injury. The association of the diagnosed NSHL with the vestibular deficiency can suggest an inner ear global deficit. In this regard, in our group of patients, 24.4% of ears with profound hearing loss associate saccular areflexia. Even more, Verbecque *et al.* suggest that the prevalence of the vestibular deficit is higher directly proportional with the degree of hearing loss [19].

There are limited methods for the quantitative assessment of the vestibular function in very small children, one of the most used tests being the cVEMPs. This is a rapid and non-invasive test with high specificity and sensitivity for the saccular lesions, appropriate for children [18].

cVEMP results vary with age, but some authors have used already this test in children with very good results, despite the fact that there are not valuable guidelines published yet [20–23]. In consequence, because there is no consensus protocol for a quantitative assessment of the parameters of the P1–N1 wave complex in children, we decided to analyze only the presence or the absence of cVEMP response for our study.

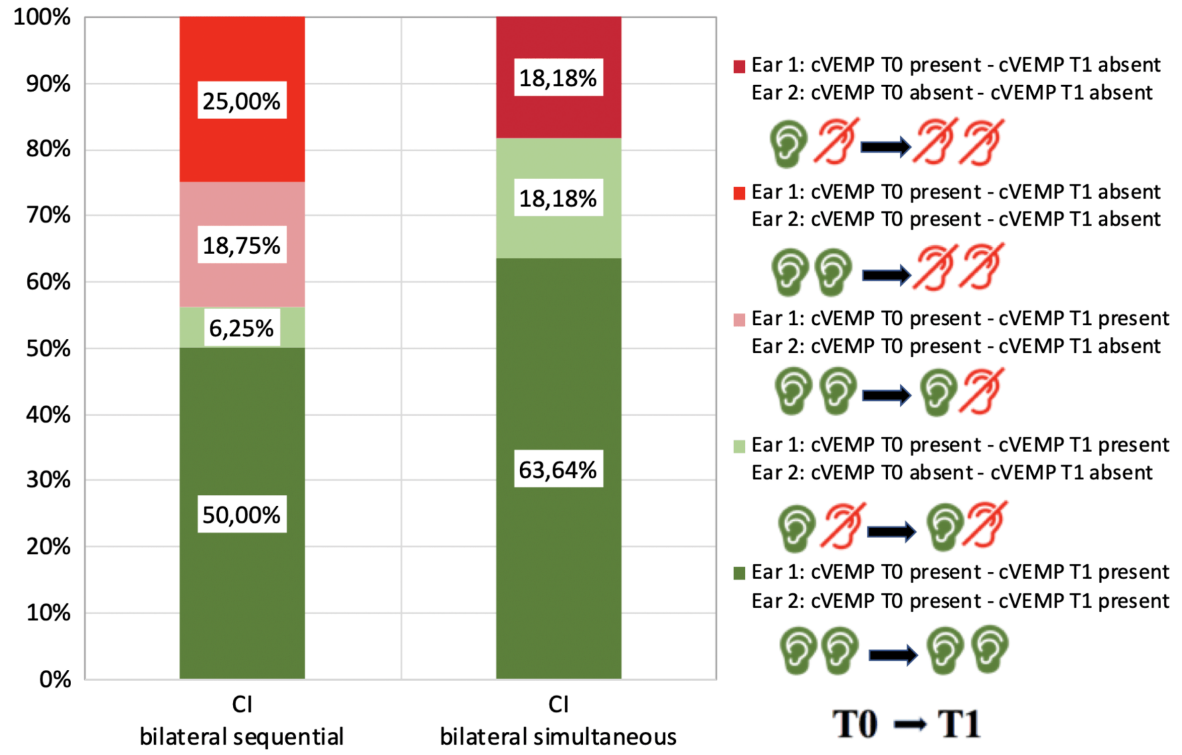

Figure 4 – ***Postoperative saccular status (T1) for bilateral sequential* versus *bilateral simultaneous cochlear implantation in children with at least one functional saccula at T0. CI: Cochlear implant; cVEMP: Cervical vestibular evoked myogenic potential.***

In our study, the bilateral saccular loss occurred in the group of bilateral sequential cochlear implantation, while in the group of simultaneous implantation, we have not identified any bilateral loss induced by the implantation. However, there are in this group some patients with bilateral saccular loss due to the unilateral damage produced by the cochlear implantation associated to a preoperative contralateral dysfunction.

Analyzing the saccular status for the group of bilateral cochlear implantation depending on the strategy of simultaneous or sequential surgery, we observe the following: more than half of our patients have conserved the saccular function on both ears after the surgery (63.64% of simultaneous bilateral CI and 50% of the sequential bilateral CI); the unilateral injury induced by cochlear implantation in patients with preoperative bilateral normal function occurred in 18.75% for sequential surgery, while 18.18% lost their unique functional ear after the simultaneous surgery; the unilateral saccular function was maintained in 6.25% after the sequential implantation and in 18.18% after the simultaneous bilateral implantation; the complete bilateral loss of the saccular function induced by surgery was observed only in the sequential CI group for 25% of patients. Randomly, we observed, probably without any clinical significance, that in the subgroup of patients with preoperative unilateral saccular function, all of them have maintained it after the sequential surgery, while those who underwent the simultaneous implantation have lost it. The most important concern for bilateral vestibular loss refers to the simultaneous cochlear implantation. However, in this particular group, we did not identify any postoperative bilateral saccular loss, but this result could be explained by the small number of our bilateral simultaneous cochlear implanted children.

In our study, at the preoperative moment (T0), the prevalence of the saccular areflexia was detected in 24.44%, while after the surgery (T1) was identified in 46.66% of the cases. We can conclude that the difference between these results indirectly indicates the group of patients that lost their saccular function for surgical reasons, the most important factor being the insertion approach. The RW insertion has the benefit that is not necessary to drill the basal turn of the cochlea. Using this approach might reduce the surgical trauma [24, 25]. González-Navarro *et al.* published a study about the correlation between the surgical technique and the postoperative vertigo in adults. The conclusion of this study is that even the RW insertion could induce more vestibular symptoms in the early postoperative period, the sensorial vestibular permanent deficit is unlikely to occur [26].

The portelectrode insertion by CO involves a risk of vestibular loss due to the drilling that may produce a mechanical and thermal aggression. In addition, the bony drilling residue can penetrate into the inner ear and produce ossifications. There are discussions about the best CO place in order to assure the access for the insertion into the *scala tympani* and, in the same time, to avoid as much as possible the permanent vestibular lesions. In this regard and due to the ambiguity of the nomenclature for the topography of the CO, which makes difficult to understand certain anatomical notions, Badr *et al.* published some landmarks for the placement of the CO in a less traumatic manner in order to guarantee the insertion of the portelectrode into the *scala tympani* and to avoid as

much as possible the vestibular damages (Figure 5). The authors sustain that the most secure place to perform the CO seems to be the intersection between B and C area (intermediate CO position – ICP) [27].

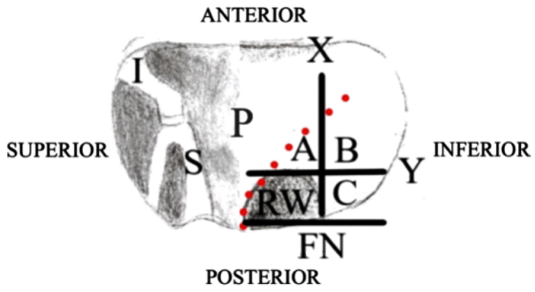

Figure 5 – ***Possible cochleostomy (CO) area for a secure portelectrode insertion. A: Anterior CO; B: Antero-inferior CO; C: Inferior CO; FN: Facial nerve; I: Incus; P: Promontorium; RW: Round window; S: Stapes. Adaptation after Badr* et al. *(2018) [27].***

The variability of the CO place is understandable and depends on the surgeon training for a specific procedure. Also, the protocol of the vestibular testing differs from one publication to another. That could explain why the studies present discordant results [19]. There are authors [28–30] sustaining that the saccular response can appear postoperative with the CI turned on. There are also publications showing that the surgical approach (CO or RW) does not influence the results of vestibular tests performed before and after cochlear implantation [5]. We present, in Table 1, a review of some studies reporting the levels of vestibular deficits [1, 5, 6, 19, 28, 31–33].

Table 1 – ***Comparative preoperative and postoperative saccular deficit in cochlear implanted patients reported by different authors***

Study	No. of enrolled patients	Preoperative saccular deficit (cVEMP)	Postoperative saccular deficit (cVEMP)
Jacot *et al.*, 2009 [31]	89/224	NA/(89)/45% (224)	51% (89)
Xu *et al.*, 2015 [29]	31	33%	34.8%
Ajalloueyan *et al.*, 2017 [32]	27	26% (7/27)	30% (8/27)
Cushing *et al.*, 2008 [5]	40	40%	No statistical significant difference
Cushing *et al.*, 2013 [6]	153/135	53%	55%
Licameli *et al.*, 2009 [33]	19	10% (2/19)	84% (16/19)
Verbecque *et al.*, 2017 (review) [19]	828	0–53%	17–84%
Psillas *et al.*, 2014 [1]	10	60%	100%
Jin *et al.*, 2006 [28]	12	50%	100% (device turned off) 66.6% (device turned on)

cVEMP: Cervical vestibular evoked myogenic potential; NA: Not available.

The vestibular impairment due to the cochlear implantation is much more important in infants, since they can benefit by bilateral devices even before walking acquisition. Jacot *et al.* report that the insertion through the RW could induce less vestibular impairment (10% of implanted children) than the CO insertion [31], results confirmed also by Todt *et al.* (13% by RW *versus* 50% by CO) [34].

Reporting our findings related to the portelectrode insertion method, we found maintained saccular function in 73.3% of the cochlear implanted ears by RW surgical approach and in 68.42% ears by CO approach. These results suggest that the RW portelectrode insertion is the recommended strategy in order to avoid the saccular vestibular impairments we already shown in previous study carried out in adults [35]. The risk of permanent vestibular deficit with affected balance abilities and other clinical manifestations should be discussed with patient before surgery [34].

If in the case of children with bilateral NSHL and monolateral congenital vestibular deficit, the principle of cochlear implantation of the deficient ear would have been applied, in order to preserve the vestibular function in at least one ear, the current study could not have been carried out because implantation on the ear with deficit does not produce any variation in the vestibular sensorial level.

Using minimally invasive surgical techniques and less traumatic devices for inner ear, we can avoid the cochlear and vestibular lesions not only for the first cochlear implantation intervention, but also for the reimplantation purposes, even these cases are not very frequent, as many studies have shown, the CI devices having a very good reliability [36–38].

Cochlear implantation, as any surgical procedure, has some risks (this study highlights the vestibular lesions), but overall is an auditory rehabilitation procedure with undeniable benefits that improve the life quality of the patients [39]. If any postoperative vestibular damage is identified, the solution of early rehabilitation should be recommended for the best and the most rapid balance recovery [40].

Conclusions

In our group of cochlear implanted children with preoperative normal saccular function, the surgery was followed by a loss of saccular function in 29.4% of cases, which confirm the significant vestibular risk of the CI surgery in pediatric population. Anyway, the benefit of CI is undeniable, but the surgical method for the port-electrode insertion could be adapted, depending on the local anatomy, in the favor of the RW approach, our study emphasizing a smaller vestibular impairment due to the surgery (26.7%) comparing with the CO approach (31.6%). However, the RW insertion and the sequential bilateral implantation are strongly recommended as the probability to induce the simultaneous bilateral vestibular loss is significantly reduced. We consider that vestibular assessment before and after cochlear implantation has a very important role for very small children, since the normal motor development depends of the normal function of sensorial vestibular structures. For those children with vestibular impairment associated to the severe to profound hearing loss or vestibular damages induced by the cochlear implantation, the knowledge of the degree of the deficit is extremely important. This can lead to an

appropriate undelayed vestibular rehabilitation treatment in order to favor the best recovery of the neuromotor skills based on the exceptional neural plasticity at this age. The vestibular training has to be done in the same time with the hearing and speech rehabilitation, process that we could define as "the early intervention in balance rehabilitation".

Conflict of interests

The authors declare that they have no conflict of interests.

Authors' contribution

Romică Sebastian Cozma and Maria Cristina Hera equally contributed to this study.

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