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CASE REPORT

Audiovestibular Findings in a Patient with Superior Semicircular Canal Dehiscence

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Abstract

Superior Semicircular Canal Dehiscence (SSCD) is a rare medical condition of the inner ear leading to auditory and vestibular symptoms. The audiovestibular symptoms exhibited by patients with SSCD can be quite similar to other otologic disorders such as Meniere's disease, patulous eustachian tube or early otosclerosis. Therefore, diagnosis and treatment of SSCD requires careful correlation of clinical presentations with radiological imaging. In this case report, pre and postoperative findings in a 59-year-old woman with a right sided SSCD are presented. Pre-operative tests showed a mild low frequency pseudo conductive hearing loss in the right ear, lower thresholds on cervical vestibular evoked myogenic potentials (cVEMP), and reduced mean vestibulo-ocular reflex (VOR) gain (0.62) on the right anterior (or superior) canal. Middle cranial fossa approach was used to close the canal dehiscence and the patient's vestibular symptoms improved immediately after the procedure. Post operatively, pure tone audiometry confirmed closure of the air bone gap, an increased threshold of cVEMP responses and the mean VOR gain decreased from 0.62 to 0.58 in the plugged right superior canal.

Keywords

Semicircular canal dehiscence, Pure tone audiometry, Pre and Post-operative assessment

Introduction

Superior Semicircular Canal Dehiscence (SSCD) was first described by Minor and colleagues in 1998 as a defect in the bone overlying the superior semicircular canal (SCC) that causes sound-induced vertigo [1, 2]. The dehiscence of the SCC creates a mobile third window in the inner ear. These changes are responsible for the increased sensitivity to bone conducted sound as well as autophony. Other common symptoms reported in patients with SSCD include hyperacusis to internal sounds (such as heartbeat, eyes moving, footsteps), varying degrees and types of hearing loss (conductive, sensorineural and mixed) [3, 4], sound -induced vertigo (Tullio phenomenon) and pressure-induced vertigo (Hennebert's sign) [3, 5]. These auditory and vestibular symptoms seen in patients with SSCD can be present together or individually and it is still not fully understood as to why signs and symptoms vary between patients [4, 2].

Factors contributing to variations in presentations of SSCD may include patency of the cochlear aqueduct and the relative compliance of the round window membrane [2]. A study utilizing temporal bone computed tomography (CT) scan found that patients with dehiscence greater than or equal to 2.5 mm exhibited both auditory and vestibular abnormalities [6,7]. Castellucci et al. (2021) [8] reported that the greater the size of the dehiscence, the poorer the air conduction thresholds and larger the air-bone gap.

The variety in clinical manifestations and the overlapping symptoms of SSCD to other possible

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otologic disorders makes differential diagnosis crucial. For example, low frequency sensorineural hearing loss can be found in Meniere's disease and conductive hearing loss could suggest otosclerosis. Accurate diagnosis of SSCD should utilize correlation of clinical symptoms with high resolution CT scan [9].

Treatment options

Currently patients with SSCD have two treatment options: conservative and surgical (middle cranial fossa (MCF) or transmastoid (TM) approach). The conservative approach includes watchful waiting and avoiding situations that trigger or worsen the symptoms. The surgical treatment is opted only when patients suffer from debilitating symptoms or who exhibit clinical manifestations.

The type of surgical approach is selected depending on the patient's anatomy, comorbidities, and the surgeon's preference and training. For example, in patients with adequate temporal bone pneumatization, TM approach provides a safe and effective alternative compared to the MCF approach [10]. Depending on whether using MCF or TM approach, different surgical techniques such as canal plugging, resurfacing and capping are used to repair the dehiscence.

Plugging the canal lumen using bone wax or fascia and bone dust hinders endolymph flow, whereas resurfacing halts any communication between brain and labyrinth by covering the dehiscence with native tissue [2]. The MCF approach provides direct visualization of the dehiscence whereas a low lying tegmen can limit access to the dehiscence while using TM approach. Nguyen et al. (2018) [11] analyzed 24 studies describing 230 patients who underwent either a MCF or TM approach and concluded that MCF approach for the repair of SSCD is associated with greater symptom resolution than TM approach. The MCF approach requires craniotomy and is associated with greater morbidity and a longer recovery time whereas the TM approach is associated with less morbidity and requires no hospital stay [12, 13]. The TM approach also confers a slightly lower risk of cerebrospinal fluid leak and intracranial complications.

Hearing and vestibular outcomes

Regardless of the surgical approach used, a successful dehiscence repair has been reported to reduce auditory and vestibular symptoms in patients with SSCD. Zhao et al. (2012) [10] reported satisfactory symptom relief and hearing preservation in patients who underwent TM procedure while the MCF approach has also been reported to be effective in alleviating debilitating vestibular issues and in improving hearing thresholds and VEMP responses [14]. A recent study published by Mekonnen et al. (2024) [15] compared pre- and post-operative symptoms in 350 SSCD repairs using MCF approach and reported significant improvements in autophony, hyperacusis, hearing, and dizziness. Although the exact mechanism underlying the resolution of the vestibular symptoms remains unknown, the correction of hypermobile fluid dynamics in the otic capsule by blocking the pressure shunt into the vestibular system is believed to be a possible explanation [16, 17].

Other analysis reported no postoperative hearing changes such as mean air conduction thresholds. Michailidou et al. (2023) [18] conducted a meta-analysis to assess hearing outcomes after TM plugging of the dehiscence and reported that the postoperative mean air conduction thresholds remained unchanged while the mean bone conduction thresholds were significantly worse. The study concluded that the TM plugging technique, although minimally worsening the inner ear threshold, is still a safe procedure for satisfactory symptom relief. More recently, Tooker et al. (2024) [19] compared postoperative outcomes in SSCD patients who underwent either MCF or TM repair, and reported that there were no significant pre or postoperative changes in median air conduction pure tone averages or air-bone gaps, or word recognition scores in both the MCF and TM groups.

There are some tradeoffs to the reported benefits of surgery in patients with SSCD. In particular, plugging has been linked to disturbance in the natural fluid dynamics in all semicircular canals. One of the first published use of vHIT in patients with SSCD was reported by Schubert et al. in 2006. Prior to surgery, they reported a reduced mean vestibulo-ocular reflex (VOR) gain on the affected side on the superior and posterior canals, and post-operatively a significantly reduced gain was still noted on the superior canal compared to other canals. Similarly, Carey et al. (2007) [17] reported reduced VOR gain only for the dehiscent canal prior to surgery, and post-surgery reduced gain was reported not only on the repaired canal but also on ipsilateral and contralateral posterior canals. The plug effect in the common crus and loss of inhibitory contribution of the plugged canals exciting the contralateral posterior canals was suggested to be the reason for these reduced VOR gains.

In the current case study, we present pre-operative and post-operative symptoms and test findings in a patient with right-sided SSCD. This case illustrates several classic signs, symptoms and test findings of SSCD. We believe that this case report will be a valuable addition to the existing literature and also be of particular interest to students of audiology and otolaryngology and to new clinicians.

Case Report

A 59-year-old woman presented to our ENT clinic with a radiological diagnosis of right sided SSCD (Figure 1). She reported the following symptoms: intermittent vertigo that is exacerbated by digital pressure over the right ear and dizziness when exposed to loud sounds and with physical exertion. She described her dizziness

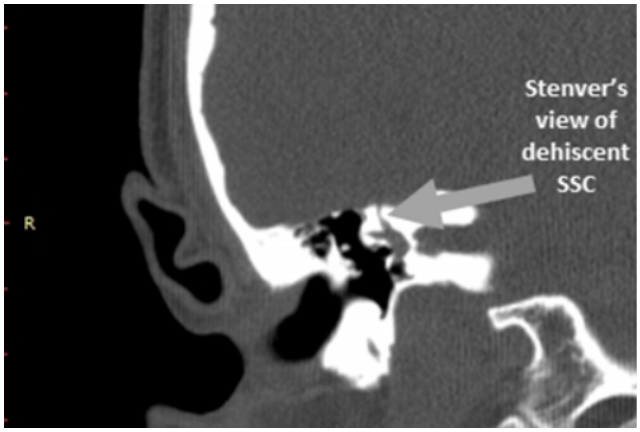

Figure 1: CT image showing a Stenvers view of the dehiscent semicircular canal.

as a constant feeling of reverberation, light-headedness and imbalance. She also reported pulsatile tinnitus, abnormal sound sensitivity, fluctuating hearing, and autophony in the right ear. Other medical conditions reported included: obesity with a body mass index (BMI) greater than 40, migraine headaches, and sleep issues.

After the initial consultation, the ENT surgeon recommended body weight management to improve overall health and monitoring of the symptoms. On the follow up ENT visit (almost 4 years after the initial consultation), the patient reported progressive worsening of her symptoms. She reported that the disability associated with her dizziness has bound her living almost exclusively on the second floor of her house. Her general health was noted to have improved with considerable weight loss (BMI: 30) achieved through dietary modifications. A combination of audiometric and vestibular tests was then performed to find clinical correlations with reported symptoms.

The surgical treatment option was considered as the symptoms had started to have debilitating effects on this patient's quality of life. The MCF approach to canal plugging and resurfacing was used to repair the canal dehiscence. This approach was selected after careful considerations of the fact that a transmastoid approach is associated with higher risk of deafness compared to MCF approach.

Audiological and Vestibular Tests

Pre-operative

Pure tone audiometry showed mild low-frequency (250 Hz, 500 Hz, and 1000 Hz) conductive hearing loss in the right ear (Table 1). The hearing thresholds in the left ear were within normal limits. Impedance audiometry showed bilateral type 'A' tympanogram suggestive of normal middle ear pressure and static compliance, with acoustic reflexes present across 500 Hz - 2 KHz test frequencies, bilaterally. The word recognition scores were 100% bilaterally. The cVEMP responses were present at both 90 nHL and 75 dB nHL in the right ear compared to absent responses at lower levels in the left ear. Video Head Impulse Test (vHIT) showed reduced mean VOR gains on Right Anterior (or Superior) (0.62) and Left Posterior (0.43) canals (RALP plane) whereas normal mean VOR gains (> 0.80) were noted for all the other canals (Figure 2). A normal VOR gain was considered to be greater than or equal to 0.80.

Post-operative

The surgical outcomes including changes in symptoms, audiometric results, cVEMP and vHIT results were evaluated one-year post-surgery. Patient reported that her vestibular symptoms improved immediately after the surgery (Table 2). She reported no complaints of dizziness or autophony in the follow up visit, however she reported ongoing right-sided pulsatile tinnitus.

Pure tone audiometry showed a decrease in air conduction thresholds by 10 dB at 250 and 500 Hz which provided closure of the pseudo air-bone gap at low frequencies in the right ear. This could suggest that the surgical repair improved the air conduction thresholds. Type 'A' tympanograms and acoustic reflexes were noted bilaterally. The word recognition scores continued to be 100% in both ears.

Surgical repair increased the cVEMP thresholds. Postoperatively, the cVEMP responses were present only at 90 dB nHL in the right ear and were absent at lower threshold levels (75 dB nHL). The mean VOR gain decreased from 0.62 to 0.58 in the plugged right superior canal. Reduction in mean VOR gains were also observed in the right posterior canal (0.88 to 0.69) and left posterior canal (0.53 to 0.43), postoperatively. Normal VOR gains were noted in both lateral canals and left superior canal.

Table 1: Pre and post-operative air conduction and bone conduction thresholds in decibels are shown here.

	Pure tone audiometry			
		250 Hz	500 Hz	1000 Hz
Air conduction	Pre	35	30	25
	Post	25	20	25
Bone conduction	Pre	10	15	10
	Post	5	15	20

Table 2: Pre and post-operative symptoms reported by the patient. *Pulsatile tinnitus continued to be reported one year after the surgical repair of the dehiscence.

Symptoms	Pre-operative	Post-operative
Autophony	Present	Absent
Abnormal sound sensitivity	Present	Absent
Pulsatile tinnitus*	Present	Present
Fluctuating hearing	Present	Absent
Tullio Phenomenon	Present	Absent
Hennebert's sign	Present	Absent
Imbalance & Light headedness	Present	Absent

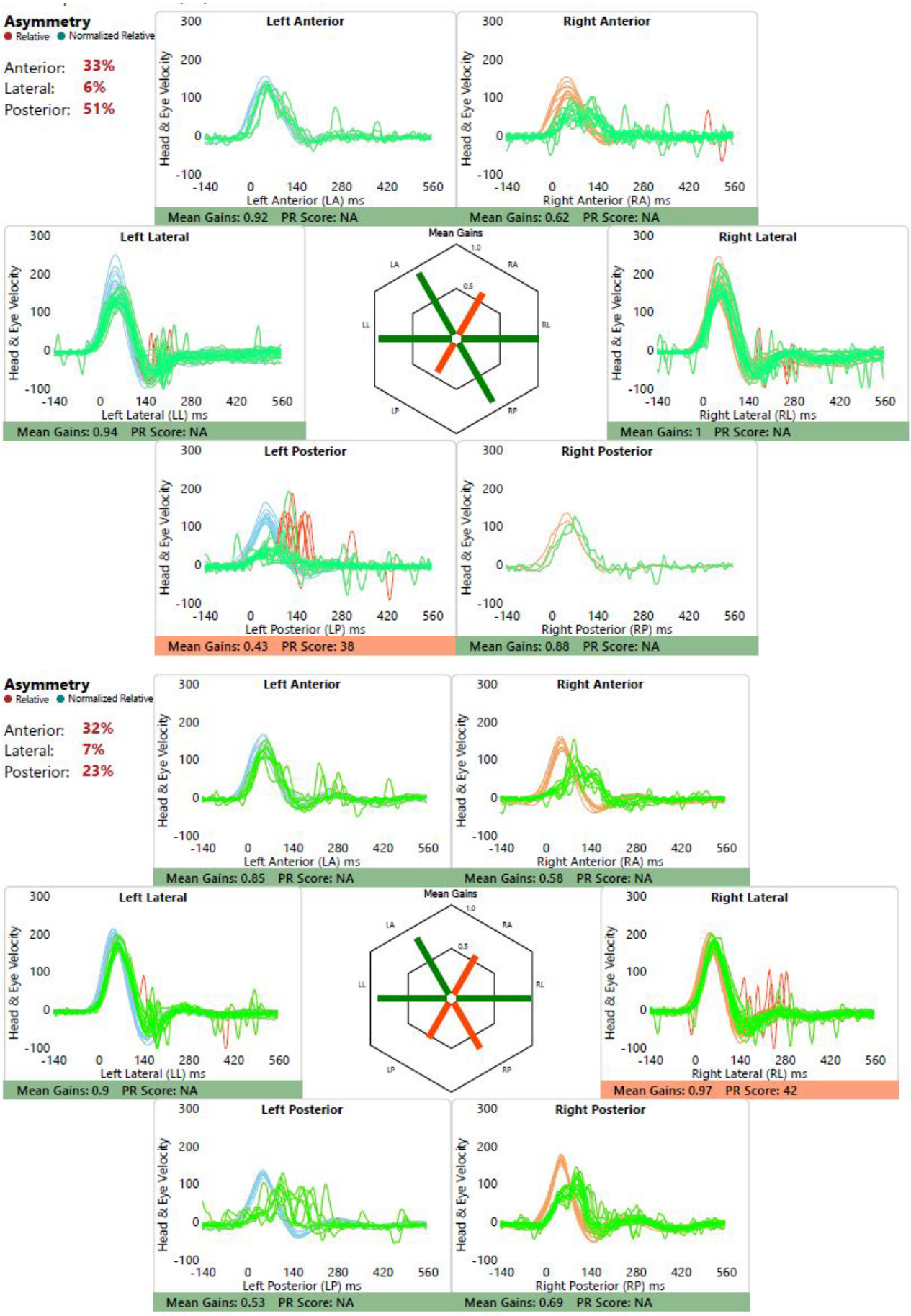

Figure 2: Pre (top) and post-operative (bottom) VOR gains obtained from all six semicircular canals.

Discussion

Audiometric and vestibular tests have been used in the detection and evaluation of dehiscence syndromes [20, 21, 22]. Zhou et al. (2007) [22] reported that VEMP is highly sensitive and specific for SSCD, possibly better than CT scan. Although CT scan can provide important diagnostic information about SSCD, it may not be sufficient for diagnosis. This is because many patients with imaging evidence of dehiscence may be asymptomatic, which reflects the protective nature of the inelastic dura in preventing pressure transmission through the bony dehiscence. Combining audiometric and vestibular tests have proven to be useful to confirm SSCD diagnosis in these instances [14, 23].

Our study illustrates how the audio vestibular test findings along with patient reported symptoms can provide guidance to surgeons regarding the treatment selection to balance the risks associated with surgery. We used pure tone audiometry, cVEMP and vHIT to evaluate the patient with SSCD reported in this case study. We then compared the pre and post-operative findings in this patient using the same tests. Pre-operatively we found a pseudo air bone gap, reduced cVEMP thresholds and reduced VOR gain isolated to the RALP plane only which was consistent with the dehiscent canal. These findings are illustrative of several classic observations reported by various authors, all within a single case.

Differential Diagnosis

SSCD has been labeled as a 'great otologic mimicker' as patients manifest symptoms similar to many other ear disorders. For example, autophony of one's own voice and sensation of 'blocked ear' would mimic SSCD as Patulous Eustachian Tube (PET). Autophony was reported by our study patient, but PET was not ruled out. We assumed that this symptom was associated with the dehiscence as she also reported Tullio phenomenon and Hennebert's sign which are classic symptoms predominantly reported in patients with SSCD. However, an objective and efficient way to diagnose PET is by performing compliance over time test using a tympanometer. During this test, the patient is asked to breathe in and out (or sniffing) through the ipsilateral nostril while plugging the other side to see the effect of tympanic membrane's compliance over time. Exaggerated changes in tympanic membrane's compliance synchronous with inhalation and exhalation is indicative of PET.

Fluctuating hearing, aural fullness and dizziness reported by SSCD patients can be misinterpreted as early signs of Meniere's disease. Distinguishing between SSCD and Meniere's disease is critical given the distinct treatment and outcomes of these two disorders. Low frequency sensorineural hearing loss is reported in Meniere's disease and SSCD, however episodic vertigo is prominent in Meniere's disease. In this case study, a low frequency conductive hearing loss was noted, which was ruled out to be a pseudo-conductive loss by the presence of acoustic reflexes and type 'A' tympanogram on the dehiscent side. Moreover, the dizziness that she reported was persistent feelings of reverberation and light-headedness rather than episodic vertigo (spinning-type) which is common in Meniere's disease. Other tests such as Electrocochleaography (ECochG) and VEMP can also provide useful diagnostic information relevant to differentiate Meniere's disease and SSCD.

Previous research has reported that on pure tone audiometry the most common findings in SSCD is a large air-bone gap in the low frequencies (250 Hz, 500 Hz, 1000 Hz), which could also be suspected as an early sign of otosclerosis. However, in such instances it is important to consider performing acoustic reflex test which is usually present in patients with SSCD and absent in cases involving otosclerosis. This is demonstrated in this case study. Even though there was a low frequency conductive hearing loss demonstrated on the pre-operative audiometry, the presence of acoustic reflexes along with type 'A' tympanogram helped us to rule out a true conductive component.

Vestibular tests can also help in the differential diagnosis of SSCD and otosclerosis. Lower than normal thresholds for cVEMP responses and elevations in the cVEMP amplitudes reported in SSCD cases can be of diagnostic importance. It is known that cVEMP responses would be mostly absent in patients with otosclerosis due to the conductive component in the middle ear [24]. A recent study using vHIT in patients with unilateral otosclerosis reported normal vHIT mean gains in the affected side, and no significant difference between the ears in the affected group [25]. In SSCD, a reduced VOR gain and catch up saccades isolated to the affected superior semicircular canal has been consistently reported [20, 21]. It could be inferred that if isolated superior canal abnormalities are demonstrated in vHIT then that could be suggestive of SSCD and patients need to be referred for further investigations.

Patients with SSCD have also been reported to have a higher mean body mass index and history of obstructive sleep apnea compared with those without dehiscence [26]. Schutt et al. (2015) [26] reported a possible causality between increased intracranial pressure and the formation of SSCD. It is interesting to note that our study patient also had obesity and sleep issues while also exhibited a variety of classic audiological and vestibular manifestations of SSCD reported in the literature. [27-29]

Post-operative improvements

Previous research in patients after dehiscence repair has reported improved hearing thresholds, improved autophony [5], normalized VEMP amplitudes and thresholds [16] and relief from sound- and pressure-induced vertigo [17]. Another study using

vHIT within one week after surgery had reported vestibular hypofunction in all ipsilateral canals and some contralateral canals as well as the emergence of compensatory saccades at an early phase [16]. Carey et al. (2007) [17] reported that the severely reduced VOR gain for plugged canals remains unchanged over time, whereas the VOR function in other canals generally, if not always, resolves after surgery.

In summary, we observed improvements in hearing thresholds, normalization of cVEMP thresholds, and instant patient-reported relief from autophony and sound and pressure induced symptoms after dehiscence repair. The air conduction thresholds improved on the repaired side and the cVEMP responses were obtained only at high intensity levels after the repair. The vHIT test was also repeated one year after the surgery and confirmed that the low mean VOR gains noted at the RALP plane persisted after the surgery. In addition, a low mean VOR gain was also observed in the right posterior canal post-operatively.

Limitations and future direction

The short follow-up periods after surgery and small sample sizes make it challenging to establish definitive associations between changes in audiovestibular function and surgical repair of SCD, especially concerning long-term vestibular function. Future studies should investigate longitudinal vestibular function in patients undergoing SSCD repair and include larger sample sizes.

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Disclosures

No conflicts of interest, financial or otherwise are declared by the author.

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