**Abstract**

**Objectives:** The goal of the present study was to examine the effects of unilateral cochlear implant (CI) surgery on each sensory organ of the balance system separately, in adult patients with bilateral severe to profound sensory neural hearing loss (SNHL).

**Methods:** We recruited 7 CI candidates aged between 18 to 70 years (four women, three men) and measured the function of the sacculus, utricle, and the three semi-circular canals (SCCs) using air conduction cervical vestibular evoked myogenic potential (ACS cVEMP), bone conducted vibration ocular VEMP (BCV oVEMP) with a mini-shaker, and the video head impulse test (vHIT) respectively on each side, pre- and post-operatively.

**Results:** Our study suggest that the otoliths are more affected by implantation than the SCCs. In 3 of 7 cases VEMP was affected by implantation and in 2 cases this change was associated with short-term dizziness.

**Discussion:** Improved vestibular testing methods now allow differential assessment of individual vestibular organ function and this may lead to a better understanding of how cochlear implantation affects the vestibular system. As many CI candidates have some vestibular function, pre-implant vestibular assessment may help to inform which side of implantation may best preserve that function where other audiology and surgical considerations are equal. Post-implant assessment with VEMP may help to predict short-term dizziness. More work with a larger sample will be needed to make the case for routine clinical assessment.

**Conclusions:** This study highlights the potential benefit of conducting multimodal vestibular function testing pre and post cochlear implantation. The otoliths appear more affected by implantation than the SCCs.

**Keywords**: Unilateral cochlear implant; Adults; ACS VEMP; BCV oVEMP; vHIT; Vestibular function.

# Introduction

Cochlear implants (CIs) are widely used to initiate or restore hearing for patients who do not get benefit from conventional hearing aids (Santos et al., 2015). The effect of severe to profound hearing impairment on quality of life is well established (e.g. Tatovic et al., 2011) and the benefits of CI to improve hearing, speech perception, language production and quality of life are well documented (for example see systematic reviews on the clinical effectiveness of CI in severely-profoundly deaf children and adults such as Forli et al., 2011 and Berrettini et al., 2011). Although fitting a CI is considered a safe procedure, opening a hole in the cochlea when inserting the electrodes has potential to damage the neighbouring vestibular organs through trauma, infection, haemorrhage, vascular changes due to inserting of the electrodes, or pathologic disruption of the endolymphatic system (Santos et al., 2015). Examples of this have been confirmed by histopathological studies (Handzel et al., 2006; Tien and Linthicum, 2002) and several studies that used clinical tests pre-and post-surgery, such as the video Head Impulse Test (vHIT) (Batuecas-Caletrio et al., 2015), and Vestibular Evoked Myogenic Potentials (VEMPs) (Basta et al., 2008; Jin et al., 2008; Licameli et al., 2009; Psillas et al., 2014; Robard et al., 2015).

Recently, the risk of vestibular damage following CI surgery has received more attention because of the possibility of bilateral implantation (Wagner et al., 2010). Several studies have evaluated the incidence of vestibular damage after CI surgery (Basta et al., 2008; Jacot et al., 2009; Jin et al., 2008). In these studies, the reported incidence of vestibular function loss (change in vestibular end-organs function as measured objectively by clinical tests) following implantation varies widely, with estimates, ranging between 23 and 100 % of patients (Robard et al., 2015). This vestibular loss seems not to be related to the aetiology of hearing loss but may come from surgical trauma or from electrical stimulation of the CI electrodes (Filipo et al., 2006). Although vestibular loss following implantation has been proven in histopathological studies (Handzel et al., 2006; Tien and Linthicum, 2002), the mechanisms to explain this damage after operation are still poorly understood (Abouzayd et al., 2016). An additional complication is that there is no correlation between the patient’s vestibular symptoms measured by the dizziness handicap inventory (DHI) questionnaire and the outcome of the objective balance tests (Abouzayd et al., 2016): Neither cVEMP, caloric nor vHIT have been found to have a good correlation with a patient’s balance symptoms. A possible explanation for the poor correlation between the outcome of these objective tests and a patient’s subjective symptoms may be that objective testing measures end organ function, whereas dizziness will depend on whether or not central compensation for vestibular dysfunction has occurred and this may not be measured directly by objective tests (Abouzayd et al., 2016). This poor correlation might also be attributed to limitations in the sensitivities of the methods and in the case of caloric testing, the low frequency nature of the caloric stimulus compared to every day vestibular excitation.

A further reason for the poor correlation of individual test results with balance symptoms may be that each test selects specific vestibular end organs. Hence untested organs could be responsible for symptoms that are not detected using just one or a few objective balance tests (Abouzayd et al., 2016). Unfortunately, it is unlikely that the five vestibular organs can be measured with a single balance test. Consequently, it has been suggested that measuring balance function following CI surgery should involve a comprehensive assessment for all vestibular end organs and, for example, should include cVEMP (saccule), oVEMP (utricle), and vHIT (all three SCCs) (Maheu et al., 2017).

VEMP is widely used in clinical practice as an objective vestibular technique for measuring the function of otolith organs. Sound evoked cVEMP is thought to predominately represent the saccular pathway function (Rosengren et al, 2010). The saccular macula is the closest vestibular organ to the cochlea, and, consequently, it is the vestibular organ most frequently impaired by CI surgery, and also the sensor most likely to be damaged following implantation, according to histological studies (Handzel et al., 2006; Tien and Linthicum, 2002). To date, the measured rate of saccular dysfunction using sound evoked cVEMP testing reported after CI surgery is between 21‒100 % (Basta et al., 2008 ; Jin et al., 2008; Krause et al., 2010; Licameli et al., 2009; Melvin et al., 2009; Robard et al., 2015; Todt et al., 2008; Xu et al., 2014). More recently, in a large systematic review and meta-analysis of 16 studies, Abouzayd et al. (2016) reported a sensitivity of 32 % for cVEMP testing to detect saccular dysfunction in symptomatic CI users. However, the 16 studies included in the review had many methodological differences, for instance, in terms of stimulus type, level of stimulation and criteria for analysing the presence of the cVEMP response. One possible explanation for changes in the response of some objective measures of vestibular end-organ function (i.e., VEMP) could be due to a change in sound transmission in the implanted ear rather than saccular disorder (Basta et al., 2008). However the general association between dizziness and VEMP responses reported in Abouzayd et al. (2016) implies that changes may occur in the vestibular system following implantation in addition to changes in the sound transmission pathway.

The evaluation of the effect of CI on utricular function has received little attention in the literature. To the best of our knowledge, only two studies have measured utricular function before and after implantation using sound evoked oVEMPs. Xu et al. (2014) reported that 82 % (18 out of 22 young patients) of children with clear AC oVEMP responses preoperatively showed utricular dysfunction in the implanted ear following surgery. Maheu et al. (2017) found that 75 % (3 out of 4) of patients showed a complete loss of ocular VEMP responses in the implanted ear postoperatively. This postoperative loss of oVEMP responses in the implanted ears may reflect damage to the utricle by implantation (Xu et al., 2014).

There is still some debate as to whether it is the type of measurement (e.g. cVEMP or oVEMP) or the stimulation method (sound or vibration) that selects whether the sacculus or utricle is being tested with VEMP (see Todd, 2014, for a review). Curthoys (2010) proposed that oVEMPs reflect contralateral utricular function and cVEMPs reflect ipsilateral saccular function. In this scheme, oVEMP and cVEMP are independent measures that can be used to differentiate responses of the saccular and utricular macula due to differential neural projections to various muscle groups and it is the response being measured rather than the type of stimulation that determines which end organ is selected. However, Todd et al. (2008) suggest that it is the type of stimulation used that selects the end organ, with 100 Hz low frequency vibration selectively stimulating the utricle and 500 Hz sound the saccule.

There does however appear to be agreement in the literature that 100 Hz vibration oVEMPs primarily reflect utricular function. Hence in the present study this was used as a test of utricular function. As the technique of measuring oVEMP in response to mini-shaker vibration is new, there is limited normative data to define the presence or absence of a response in the literature. Therefore in our study we measured normative data for oVEMPs to vibration (via mini-shaker) on a small sample of healthy subjects, for comparison to clinical measurements pre and post implantation.

vHIT measurement allows relatively fast assessment of semi-circular canal function e.g. Migliaccio et al. (2005) and was initially introduced for horizontal canal measurement. Reported deterioration in the function of horizontal SCCs after CI surgery varies from 19 % (Todt et al., 2008) to 72.4 % (Robard et al., 2015). The variability could be attributed to differences in the test techniques used for recording and/or the criteria (low cut-off value) used to differentiate between normal and abnormal responses. To date, relatively few studies have evaluated the function of both vertical and horizontal SCCs before and after implantation, using vHIT or 3D HIT. e.g. Migliaccio et al. (2005), Maheu et al. (2017), and Melvin et al. (2009) tested each of the three canals. e.g. Migliaccio et al. (2005) found a significant decrease in vestibulo-ocular reflex (VOR) gain for all three canals using vHIT on the side of the implant for one symptomatic patient (9 %) following CI surgery. Melvin et al. (2009) found one patient (3.6 %) with a significant drop in vestibular function in all three SCCs (from normal, to severe, to profound vestibular dysfunction on the 3D HIT). Maheu et al. (2017) did not report any loss of function in any patient on the vHIT and suggested that the function of SCCs is not affected by CI surgery. Further studies have tested only specific canals. e.g. Basta et al. (2008) failed to find any patient with a loss of vertical canal function on the vHIT following implantation. Overall there are conflicting findings in the literature regarding the effects of CI on vHIT measures of canal function.

To the best of our knowledge, only two recent studies have measured each of the five sensory vestibular organs separately using vHIT, and VEMPs and none used vibration elicited VEMP. Maheu et al. (2017) measured the impact of unilateral implantation on balance function using vHIT, cVEMP and oVEMP, and showed that 75 % (3 out of 4) of patients showed a complete loss of cervical and ocular VEMP responses in the implanted ear following surgery. None of the 4 patients reported a change in vHIT, which suggests that the function of SCCs was not affected by CI surgery. However, this study has some limitations, which could affect its validity to determine the effect of implantation on balance function: only 4 patients were recruited and no reason for this small sample size is given. In addition, the authors did not mention any of the stimulus parameters, e.g. stimulus type, intensity, and duration, that were used to elicit cVEMP and oVEMP in their study and the mean delay between CI surgery and the vestibular tests was not stated.

Because of the close anatomical connection between the cochlea and the vestibulum, it is probable that patients with severe to profound Sensori-neural hearing loss (SNHL) may exhibit abnormalities in balance function. A review conducted by Santos et al. (2015) showed that almost 70 % of children with SNHL have vestibular loss, and 20-40 % of them have severe bilateral vestibular dysfunction. Deafness is associated with vestibular dysfunction in several aetiologies, such as Usher syndrome and malformations in the inner ear (Thierry et al., 2015) which can result in pre-implantation vestibular disorders. In order to assess if there is an effect of CI surgery on balance function, and rule out pre-implantation disorders, balance testing should be performed both before and after surgery.

The goal of the present study was to examine the effects of CI surgery on each sensory organ of the balance system separately, in adult patients with bilateral severe to profound SNHL undergoing unilateral implantation. Specifically, the function of the sacculus, utricle and the three SCCs was assessed using ACS cVEMP, BCV oVEMP with a mini-shaker, and vHIT respectively in each ear, both pre- and post-operatively.

# Methods

The methods used in this study with hearing impaired subjects were approved by both the Human Experiment Safety and Ethics Committee of the University of Southampton’s Institute of Sound and Vibration Research (ISVR) and the NHS Ethics Integrated Research Application System (REC reference: 14/WA/1015 and IRAS project ID: 156658) before the research commenced. Ethics approval for the normative study of oVEMP in response to mini-shaker vibration was also obtained from the University of Southampton Ethics Committee. Our study was independent of the clinical management of patients, so although the results of the testing were passed to the clinical team, the results were not used to inform the clinical management of the patients.

## 2.1 Study population

### Normative study of oVEMP to vibration with a mini-shaker

10 subjects (6 male and 4 female) aged between 18 and 45 years, with a mean age of 26 years, were recruited in the study. Screening questionnaires were used to rule out balance problems and significant problems in vision that could not be reversed with eyeglasses or contact lenses. It was also ensured that subjects had normal hearing using PTA, and all subjects had pure-tone thresholds of around or better than 20 dB HL (hearing level). oVEMP responses to vibration (mini-shaker) were recorded from 10 ears from the 10 normal subjects. The normal subjects had similar, normal pure-tone average from both ears and it was decided to collect data from their right ears.

### Study group (CI patients)

This study was conducted on seven deaf CI recipients (four women, three men) aged between 45 to 70 years, with a mean age of 57 years, who were met the criteria of the present study and were operated on at the University of Southampton Auditory Implant Service (USAIS) between 2015 and 2017. All CI recipients included in the study were implanted using a similar surgical approach-known as the extended round window approach ([O'Connell](https://www.ncbi.nlm.nih.gov/pubmed/?term=O'Connell%20BP%5BAuthor%5D&cauthor=true&cauthor_uid=28894813) et al., 2016).

The adult patients were selected for implantation based on NHS criteria ([NICE TAG 166 Guidelines](https://www.nice.org.uk/Guidance/TA166)). The main criterion for implantation was having severe to profound SNHL with no benefit from conventional hearing aids. Patients who had back/ neck pain, visual problems, outer and middle ear pathology (e.g. excessive wax, infection, ear discharge or bleeding), and allergy to alcohol swab were excluded from the study. All patients underwent computed tomography (CT) of peterous bones and Magnetic resonance imaging (MRI) scan of the internal auditory meatus (IAMS), but in all cases this testing found no abnormalities. The majority of the patients (5) included in the study had acquired SNHL from unknown causes. Two patients had congenital deafness due to maternal rubella and bacterial meningitis, respectively. The complete patients’ information is shown in **Table 1.** All patients gave their written informed consent. The medical records for each patient were obtained from USAIS in order to document any history of balance problems or any relevant balance testing results, to compare them with the results of this study. The patient’s evaluation of the vestibular function was collected from each patient before and after CI surgery, using a modified version of the Vestibular Rehabilitation Benefit Questionnaire (VRBQ) ([http://www.isvr.soton.ac.uk/audiology/vrbq.htm)](http://www.isvr.soton.ac.uk/audiology/vrbq.htm%29). Evaluation of self-reported balance function was carried out one day to six weeks before and two to three months after implantation.

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| **Patient number**  | **Sex**  | **Age at onset of hearing loss**  | **Aetiology of hearing loss**  | **CI device**  | **Surgeon symbol**  |
| **1**  | F  | 52 years  | Unknown  | Cochlear CI512  | B  |
| **2**  | M  | 54 years  | Unknown  | AB Mid Scala  | A  |
| **3**  | M  | 54 years  | Unknown  | Cochlear CI512  | B  |
| **4**  | F  | Congenital  | Maternal Rubella  | Cochlear CI512  | C  |
| **5**  | M  | 6 years  | Unknown  | Cochlear CI512  | B  |
| **6**  | F  | Early childhood  | Bacterial meningitis  | AB HiRes Ultra CI  | A  |
| **7**  | F  | 39 years  | Unknown  | Cochlear CI512  | C  |

**Table 1: Demographic data for seven implanted patients who took part in this study. F denotes female, M denotes male, and CI denotes cochlear implant. A, B, and C represent the surgeons who carried out the surgery.**

## 2.2 Stimuli and Apparatus

### VEMP testing

Cervical and ocular VEMPs were measured using the Bio-Logic Auditory Evoked Potentials system (AEP version 6.2.0) monaurally through headphones (TDH-39P) and a mini-shaker vibrotactile perception meter (VPM). For AC cervical VEMP, stimuli were 500 Hz tone-burst stimuli with 1-cycle rise/fall, and 2-cycle plateau at sound intensity level of 90 dB nHL. 10-1500 Hz band-pass filters were applied to the data. Each recording required 150 repeats of a 0.008 s tone pip, (4 cycles with a cycle duration of 0.002 s), making the total stimulus duration for one recording 30 s. The repetition rate (5 Hz) was constant, so the total recording time for 150 epochs was 30 s (time window of 0.2 s × 150 epochs). The output from the Bio-Logic AEP system (version 6.2.0) was routed monaurally through headphones (TDH-39P). Four recordings (2 recordings for each ear) were repeated to give a total exposure time of 120 s.

Vibration ocular VEMPs (via a mini-shaker) were measured using 0.4 s tone bursts at 100 Hz with an rms (root mean square) level of 1 ms-2 (metre per second squared) placed on the mastoid bone directly behind the auricle. For each vibration, 150 epochs of 0.4 s were generated (60 s exposure). Four recordings were repeated to give a total exposure time of 240 s. The output from the Bio-Logic AEP system (version 6.2.0) was routed into the VPM and the acceleration signal from the mini-shaker was measured on an oscilloscope. The oscilloscope was calibrated to target a peak-to-peak value of 5.2 volts when the measurement was made on the patient, in order to obtain the target acceleration of 10 ms-2 rms (5.2 V peak to peak = 14 ms-2 or 10 ms-2 rms). The mini-shaker was applied to the head with a force of 10 N (Newtons), as indicated on a calibrated force meter.

### vHIT testing

vHIT was measured by the ICS Impulse System manufactured by GN Otometrics (2011). A laptop running OTOsuite software was connected to a pair of light-weight goggles that were firmly fixed to the subject’s head. The goggles contained both a high-speed video oculography camera that measured the subject’s eye movement and a gyroscope that measured the subject’s head velocity, as described in MacDougall et al. (2009). The eye position camera analysed the eye movement with a sampling rate of up to 250 Hz (MacDougall et al., 2009). The system measured the VOR of each SCC individually at high frequencies. The gain of the VOR is defined as the ratio of compensatory eye velocity to head impulse velocity, which is around one (Alhabib & Saliba, 2017). VOR gains which are significantly less than one indicate reduced SCC function (Curthoys & Manzari, 2017).

## 2.3 Procedures

### For each patient, the test order of cVEMP, oVEMP and vHIT was randomised. All procedures were carried out in the same session with 10 minutes interval between individual tests.

### cVEMP recording

VEMPs from the sternocleidomastoid (SCM) muscle were recorded ipsilaterally while subjects were seated upright on a chair with their chin turned over the contralateral shoulder to tense the SCM muscle. The electromyographic (EMG) activity of the SCM muscle was recorded using surface electrodes placed on the muscle: active on the belly of the ipsilateral SCM muscle, and reference on the upper sternum of the test side. A ground electrode was placed on lower forehead. The impedance of the electrodes was kept below 10 KΩ. The EMG activity of the SCM muscle was visually monitored on an oscilloscope and kept between 80 and 100 mV.

### oVEMP recording

VEMPs from the ocular muscles were recorded contralateral to the stimulated ear, in response to vibration. The responses were recorded by placing five electrodes on the face. The two active pairs of electrodes were placed on the orbital margin below the centre of the eye and referred to two reference pairs of electrodes approximately 15-30 mm below, on the cheek, and the ground electrode low on the forehead. The impedance of the electrodes was maintained below 10 KΩ. Patients sat on a chair and were asked to look up at a target located 2 m away, with an elevation of 25 degrees, whilst vibrations were applied to patient’s tested ear via the mini-shaker at the mastoid process (behind the pinna of the ear). The patients were tested with the same angle of elevation and with rest breaks in order to allow the patients to blink.

### vHIT

Patients were instructed to stare at a fixation dot located at about 1.20 m distance. About 20 head impulses in the planes of the horizontal and vertical canals were manually delivered by the experimenter with unpredictable timing and direction.

To test the lateral SCCs, the tester rotated the patient’s head horizontally in an abrupt, brief and unpredictable manner at a small angle and in a random direction (right or left). Peak head velocity of the horizontal impulses ranged from 100 to 250°/s (acceleration 1000–2500°/s2, amplitude 10–20°) (GN Otometrics, 2011).

To test the anterior and posterior SCCs, the head rotations were delivered in the planes of the vertical canals – left anterior-right posterior (LARP) and right anterior-left posterior (RALP). These canals lie in planes, which are approximately 45° to the sagittal plane of the head. The person's head was positioned about 30–40° turned to the left or right with respect to their body so that the targeted vertical canal plane was approximately aligned with the body sagittal plane, then diagonal head movements were delivered in the plane of the vertical canals, while gaze was directed at the fixed target. The tester placed one hand (dominant) on the top of the patient’s head and the other beneath the chin in order to control the direction of rotation. The patient’s head was rotated vertically via the dominant hand in an abrupt, brief and unpredictable manner with a small angle (about 10-20 degrees), while the patient attempted to maintain fixation on the fixed target. Peak head velocity of the vertical impulses ranged from 50 to 250°/s (acceleration 750–5000°/s2, amplitude 10–20°) (GN Otometrics, 2011). The direction of the head movement determines which canal of the pair is activated: so for example, downward head impulses stimulate the left/right anterior canal and the upward ones stimulate the right/left posterior canal (GN Otometrics, 2011).

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## 2.4 Response analysis

### cVEMP

The presence of a cVEMP response was visually evaluated based on specific criteria. In our work, cVEMP responses were defined as present if the cVEMP was a reproducible biphasic waveform with a positive (p1) peak followed by a negative (n1) peak, the inter-amplitude (p1-n1) of the response was between 20 and 150 μV, based on the criteria presented at the Balance Interest Group of the British Society of Audiology (2012), the waveform should be larger than the rest of the response in the overall average and the acceptable latency range of p1 was 13.9-19.2 ms and n1 was 22.9-30.3 ms. These latency values were determined from a recent study (Blakley and Wong, 2015) of cVEMP evoked by 500 Hz tone-bursts in 48 adults (23-64 years) with no history of hearing and balance problems. If any of these criteria were not met, then the cVEMP response was judged to be absent. Here all the criteria for response presence have been met, so the response is judged to be present.

### oVEMP

The normative range for oVEMP parameters to vibration with a mini-shaker at intensity of 90 dB nHL was explored in normal hearing subjects in the present study. **Table 2** shows 95 % confidence intervals for the mean (mean ± 1.96×SD) of amplitude (uV), and latencies P1-N1 (ms) of oVEMP at 90 dB nHL in 10 normal hearing subjects. Test results exceeding the 95 % confidence intervals of the mean for oVEMP parameters of the normative data (amplitude, and peak latencies) were considered abnormal in the study group (CI patients). Thus, oVEMP responses were accepted as present if the oVEMP waveform was reproducible, biphasic with a positive (P1) peak followed by a negative (N1) peak, peak-to-peak amplitude and peak latencies should be within the 95 % range shown in Table 2.

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| **oVEMP parameters**  | **Lower 95 % range**  | **Upper 95 % range**  |
| P1-N1 amplitude (uV)  | 1.02  | 3.89  |
| P1 latency (ms)  | 15.52  | 26.38  |
| N1 latency (ms)  | 19.92  | 32.48  |

**Table 2: Normative oVEMP data at 90 dB nHL: 95% confidence intervals of the mean (mean ± 1.96×SD) of P1-N1 amplitude (uV), and latencies P1-N1, of oVEMP in 10 healthy subjects.**

### vHIT

VOR gain (eye velocity/head velocity) was calculated automatically by a software package which divides the area under the curve of eye velocity by the area under the curve of head velocity (Janky and Givens, 2015). The VOR gain was considered normal if it was >0.85 for the horizontal canal, and >0.65 for the vertical canals, based on the manufacturer’s suggestion of a low cut-off VOR gain (GN Otometrics, 2011), although it is not clear which peer reviewed research journals these normative VOR values are based on (Bell et al., 2015).

# Histories and testing results for individual cases

Note: Both cVEMP and oVEMP responses are categorized as either present or absent based on specific criteria for cVEMP and normative data for oVEMP, whereas vHIT is categorized as either normal or abnormal based on the pathological cut-off VOR gain (>0.85 for the horizontal canal, and >0.65 for the vertical canals) according to the manufacturer’s suggestion of a low cut-off VOR gain (GN Otometrics. 2011).

**Patient 1**

**History** A 66-year-old woman had had bilateral severe to profound SNHL for 14 years. The cause of hearing loss was unknown. She reported that her balance was generally good. She underwent CI surgery in the right ear. She reported that her balance was generally good, and she did not notice any balance problems after the surgery, except one accident, which happened within 5 weeks after the surgery. While she was riding her bicycle, she lost her balance and fell off her bicycle. Although she did not notice a change in balance function following surgery, this accident could be consistent with a short-term effect on balance function and it highlights the impact that balance dysfunction can have. Her balance was objectively assessed one month before and three months after the CI surgery, using vHIT, AC cVEMP and vibration oVEMP. Vestibular investigations before and after the surgery revealed the following:

**Balance status before the CI surgery**

*vHIT*: the VOR gain in both ears was within the normal range for all canals.

*ACS cVEMP*: Present in both ears.

*BCV oVEMP*: Present in both ears.

**Balance status after the CI surgery**

*vHIT*: The VOR gain in both ears was within the normal range for all canals.

*ACS cVEMP*: Absent in the right ear (implanted ear) and present in the left ear.

*BCV oVEMP*: Present in both ears.

**Summary of case 1**

This patient was manifesting normal balance function before the implant. After the implant, the saccular function in the right implanted ear for this patient was affected, but the left saccular function (non-implanted ear), SCCs and utricle were unaffected.

**Patient 2**

**History** A 67-year-old man had progressive bilateral hearing loss, which had progressed to severe to profound SNHL. The cause of hearing loss was unknown. He reported that he had a minor balance problem before the implantation, as he suffered from unsteadiness when listening to loud sounds and this lasted for a few minutes and sometimes he felt dizzy when standing up after lying down. He had been implanted in the left ear. He did not notice any difference in his balance after being implanted. In addition, his balance was objectively assessed one day before and three months after the CI surgery, using vHIT, AC cVEMP and vibration oVEMP. Vestibular investigations before and after the surgery revealed the following:

**Balance status before the CI surgery**

*vHIT*: the VOR gain in both ears was within the normal range for all canals.

*ACS cVEMP*: Present in both ears. Some aspects of the patient’s history seemed indicative of superior semi-circular canal dehiscence (SCC dehiscence), so the cVEMP threshold was measured in both ears on the same day as the testing session. The cVEMP threshold went down to 85 dB nHL in both ears, so the results did not suggest a diagnosis of SCC dehiscence based on this test.

*BCV oVEMP*: Present in both ears.

**Balance status after the CI surgery**

*vHIT*: The VOR gain in both ears was within the normal range for all canals (horizontal and vertical).

*ACS cVEMP*: Absent in the left ear (implanted ear) and present in the right ear.

*BCV oVEMP*: Present in both ears.

**Summary of case 2**

This patient was manifesting normal balance function before the implant**.** Following implantation, the saccular function in the left implanted ear for this patient was affected, but the right saccular function (non-implanted ear), SCCs and utricle were unaffected. This patient reported in the self-reported questionnaire that he suffered from unsteadiness after exposure to loud sound and sometimes felt dizzy on standing up after lying down. This could be an indication of SCC dehiscence. However, results of cVEMP testing were not consistent with vestibular loss due to SCC dehiscence.

**Patient 3**

**History** A 66-year-old man had had bilateral profound SNHL for over 12 years. The cause of hearing loss was unknown but could be due to significant noise exposure during his working life. He reported that he had normal balance and he did not suffer from problems in his balance system. He was implanted in the left ear. He did not notice any difference in his balance after being implanted. In addition, his balance function was objectively assessed one month before and two months after the CI surgery using vHIT, AC cVEMP and vibration oVEMP. Vestibular investigations before and after the surgery revealed the following:

**Balance status before the CI surgery**

*vHIT*: The VOR gain in both ears was within the normal range for all canals (horizontal and vertical).

*ACS cVEMP*: Missing data. This could be due to difficulty in identifying the SCM muscle, and this was because of thick neck tissue for this patient.

*BCV oVEMP*: Present in both ears.

**Balance status after the CI surgery**

*vHIT*: the VOR gain in both ears was within the normal range for all canals.

*ACS cVEMP*: Absent in both ears. This could be due to difficulty in identifying the SCM muscle, due to the thick neck tissue of this patient.

*BCV oVEMP*: Present in both ears.

**Summary of case 3**

This patient showed bilateral normal function of both utricle and SCCs, and saccular dysfunction in both sides before the implant. Although the cVEMP results were consistent pre-post CI surgery the absence of cVEMP responses could be due to difficulty in identifying the SCM muscle, which was due to the thick neck tissue for this patient. After the implant, there was no effect of CI surgery on the other vestibular organs for this patient.

**Patient 4**

**History** A 49-year-old woman had bilateral congenital severe to profound SNHL, which was attributed to maternal rubella. She reported that her balance was generally good. She was implanted in the right ear. She had no significant balance problems immediately after surgery but had an episode of imbalance two weeks later, which could be consistent with short-term dizziness. Her balance was objectively assessed two weeks before and three months after the CI surgery, using vHIT, AC cVEMP and vibration oVEMP. Vestibular investigations before and after the surgery revealed the following:

**Balance status before the CI surgery**

*vHIT*: The VOR gain in both ears was within the normal range for all canals.

*ACS cVEMP*: Present in both ears.

*BCV oVEMP*: Absent in both ears.

**Balance status after the CI surgery**

*vHIT*: the VOR gain in both ears was within the normal range for all canals.

*ACS cVEMP*: Absent in the right ear (implanted ear) and present in the left ear.

*BCV oVEMP*: Absent in both ears.

**Summary of case 4**

Pre-implant this patient was manifesting abnormal utricular function in both sides, and normal function of both saccule and SCCs in both ears. After the implant, the saccular function in the right implanted ear for this patient was affected but the left saccular function (non-implanted ear), and SCCs were unaffected. This change in cVEMP result post implantation might be linked to the short-term dizziness reported by this patient.

**Patient 5**

**History** A 43-year-old man had congenital bilateral profound SNHL, possibly due to maternal Rubella. He was implanted in the left ear. He reported that he had started to suffer from balance problems in the last 10-12 years. He reported that he leant to the right side during walking most of the time, and that he did not notice any difference in his balance after being implanted. In addition, his balance was objectively assessed six weeks before and three months after the CI surgery using vHIT, AC cVEMP and vibration oVEMP. Vestibular investigations before and after the surgery revealed the following:

**Balance status before the CI surgery**

*vHIT*: Missing data. This could be due to a double vision problem (as the patient had reported).

*ACS cVEMP*: Present in right ear, but absent in left ear.

*BCV oVEMP*: Missing data. This could be due to his double vision problem.

**Balance status after the CI surgery**

*vHIT*: The VOR gain in both ears was abnormal in all canals.

*ACS cVEMP*: Present in right ear, but absent in left ear (implanted ear).

*BCV oVEMP*: Absent from both ears.

**Summary of case 5**

This patient manifested abnormal function of utricles, bilateral SCCs, and left side saccule, and normal right side saccule before the implant. So, he had only a small vestibular function before the implantation, which highlights the importance of performing the CI surgery on the side with poorer balance function. The patient had double vision, which may affect the performance of vHIT, and vibration evoked oVEMP, as both tests require the patient to stare at a target during testing. For example, the patient struggled to focus on the target due to his double vision, so he saw two targets on the wall instead of one. After the implant, there was no effect of CI surgery on the saccular function in the non-implanted ear (right) for this patient.

**Patient 6**

**History** A 55-year-old woman had had longstanding bilateral severe to profound hearing loss since early childhood, caused by bacterial meningitis which completely destroyed the balance and hearing parts of the ear. She reported that she suffered from severe balance problems, and that the symptoms were undoubtedly worse in the dark. She was diagnosed with bilateral peripheral vestibular hypofunction. She underwent CI surgery in the right ear. Her balance was objectively assessed two days before and two months after the CI surgery, using vHIT, cVEMP elicited by air conduction sound and oVEMP evoked by vibration. Vestibular investigations before and after the surgery revealed the following:

**Balance status before the CI surgery**

*vHIT*: The VOR gain in both ears was abnormal for all canals (horizontal and vertical).

*ACS cVEMP*: Absent in both ears.

*BCV oVEMP*: Absent in both ears.

**Balance status after the CI surgery**

*vHIT*: The VOR gain in both ears was abnormal for all canals (horizontal and vertical).

*ACS cVEMP*: Absent in both ears.

*BCV oVEMP*: Absent in both ears to 100 Hz tone-burst stimulus.

**Summary of case 6**

This patient was manifesting abnormal bilateral function of SCCs, utricle and saccule prior to implantation. Nothing changed in her balance results after the implant. This woman was very fit and active, which was slightly surprising given her vestibular hypofunction.

**Patient 7**

**History** A 54-year-old woman had noticed a steady decline in her hearing approximately 15 years ago. The cause of her hearing loss was unknown. She reported that her balance was generally good, and she had not previously suffered from balance problems. She underwent CI surgery in the left ear. She was off balance for a couple of days following surgery (short-term vestibular disorders) but otherwise she had made an uneventful recovery. Her balance was objectively assessed four months before and two months after the CI surgery using vHIT, AC cVEMP sound and vibration oVEMP. Vestibular investigations before and after the surgery revealed the following:

**Balance status before the CI surgery**

*vHIT*: The VOR gain in both ears was within the normal range for all canals (horizontal and vertical).

*ACS cVEMP:* Present in both ears.

*BCV oVEMP*: present in left ear and absent from right ear.

**Balance status after the CI surgery**

*vHIT*: The VOR gain in both ears was within the normal range for all canals (horizontal and vertical).

*ACS cVEMP*: Present in both ears.

*BCV oVEMP*: absent in both ears.

**Summary of case 7**

This patient was manifesting normal function of bilateral SCCs, saccules, left side utricle, and abnormal right side utricle before the implant. After the implant, the utricular function in the left implanted ear for this patient was affected, which might be consistent with the short-term feeling of being off-balance following surgery. However, the saccular and SCCs function in both ears were unaffected after CI surgery.

**Overall Summary of Results**

**Table 3** shows a summary of the changes in balance function for the seven deaf patients from pre to post-operatively in the implanted and non-implanted ears. 50 % of deaf patients (i.e. three out of six, excluding patient 3 with missing cVEMP data), and 16 % of deaf patients (1 out of six, excluding patient 5 with missing oVEMP data) showed loss in saccular and ocular responses, respectively, following implantation. The cVEMP response was the most affected by surgery, followed by the oVEMP response, whereas the vHIT response yielded no change following CI surgery.



**Table 3: Results of balance tests performed before and after the CI surgery for all seven patients, for the implanted and non-implanted ears. N refers to normal; A (grey shaded area) refers to abnormal; NA refers to not available (missing data), and a highlighted box with a thick black line represents a change in vestibular function pre- and post-CI surgery on the implanted side. Normal VEMP means a response was present. Normal vHIT means VOR gain >0.85 for horizontal canal and >0.65 for vertical canals.**

# Discussion

The aim of the present study was to measure the effect of unilateral CI surgery on each vestibular organ separately by comparing function before and after implantation. It should first be mentioned that this study had some missing data for two patients. Patient 3 had thick neck tissue, which caused difficulty in identifying the SCM muscle for cVEMP testing. Patient 5 had double vision, which affected the performance of vHIT, and vibration evoked oVEMP, as both tests required the patient to stare at a target during testing.

Table 3 shows a summary of the results of all balance tests performed before and after the CI surgery for all seven patients. For patients 1, 2 and 4, the cVEMP response was altered in the implanted ear, suggesting unilateral saccular dysfunction. Patient 7 only had a change in utricular function in the implanted ear as indicated by the vibration oVEMP test. Unfortunately, this patient received her implant in the L ear with normal utricular function. Pre-implant she had abnormal utricular function on the R, so ended up with bilateral utricular abnormality. It should be noted that the testing carried out was for research only and was not used to inform clinical decisions such as side of implantation. The other patients (3, 5, and 6) all had some vestibular dysfunction preoperatively, but showed no change in their balance function postoperatively. This could be related to their balance status prior to implantation: patient 6 had congenital deafness with poor balance function bilaterally before implantation, patient 3 had missing cVEMP data due to thick neck tissue, and patient 5 had missing oVEMP and vHIT data due to double vision; thus, the effect of CI surgery was not evident in these deaf patients.

The results of testing for patient 6 appear to demonstrate the sensitivity of the applied testing methods: the patient was known to have congenital bilateral hypofunction and gave a history consistent with this. All test results (semi-circular canals with vHIT, saccule with sound evoked cVEMP and utricle with vibration evoked oVEMP) were abnormal for this subject bilaterally.

Variability in the results of objective balance tests before and after the surgery can be attributed to several factors, such as cause of deafness; type of CI device, and the occurrence of trauma to the inner ear in operations by different surgeons (see **Table 1**) as well as the reliability of vestibular testing approaches. Although statistical analysis of the results was not performed due to the small sample size, the results of the current study suggest that cVEMP and oVEMP testing should could be very informative if conducted prior to implantation. 50 % of deaf patients (i.e. three out of six, excluding patient 3 with missing cVEMP data), and 16 % of deaf patients (1 out of six, excluding patient 5 with missing oVEMP data) showed loss in saccular and ocular responses, respectively, following implantation. The cVEMP response was the most affected by surgery, followed by the oVEMP response, whereas the vHIT response yielded no change following CI surgery. Those results were consistent with those of Robard et al. (2015) and Maheu et al. (2017), who revealed a significant effect of CI surgery on cervical and ocular VEMP responses. Those results were also in agreement with those of histopathological studies, which showed that the saccule is the most affected vestibular organ following implantation, followed by the utricle, whereas the SCCs are the least affected by CI surgery (Handzel et al., 2006; Tien and Linthicum, 2002). As can be seen in **Table 3**, none of the deaf patients had any change in the vHIT outcome, showing that the SCCs’ function was not affected by implantation in the patients. A further study with a larger sample of patients is needed to confirm these findings.

One patient (7) in the current study had received the implant on the ear with better balance function, ending up with bilateral utricular dysfunction. This result highlights the importance of performing a comprehensive balance assessment before unilateral implantation. Evidence of abnormal vestibular function on one side could influence the decision regarding which ear to implant. This will need to be weighed against other factors in the decision such as surgical considerations, audiological differences and patient preference (UK Cochlear Implant Study Group, 2004), but pre-implant assessment could help to avoid implanting an ear with good vestibular function over one with poor function. Generally, only caloric testing is used as a pre-implant assessment currently, so saccular and utricular function is not considered.

In the present study, there appeared to be an association between short-term dizziness and postoperative changes in VEMP responses in CI recipients, although the sample size is low. However, not all the implanted patients who showed loss of saccular or utricular function also suffered from dizziness postoperatively. The incidence of dizziness in the deaf patients was low and short-lived following implantation. Only three of the deaf patients (1, 4, and 7) reported some dizziness for a couple of days or weeks following surgery. (It should be noted that in the current study we relied on self-report of balance disorder using the VRBQ. Future work could include measurements of functional balance). In all these patients, otolith function was affected by implantation. This appears to be consistent with short-term dizziness resulting from a change in otolith function that is then compensated for. The short term dizziness associated with a change in VEMP responses also suggests that the VEMP response indicates underlying otolith function and is not simply altered due to a change in sound transmission from implantation.

Three patients (2, 5, and 6) had abnormal vestibular function prior to implantation, but reported no change in their balance problems after implantation. For CI candidates with pre-operative balance problems, the importance of preserving residual function should be considered. Also knowing where vestibular function has been altered by CI may help to target vestibular rehabilitation for patients who may be at risk of post-surgical dizziness. In general the patients in this study showed a good compensation for their balance deficit following implantation, indicating that CI surgery did not affect the quality of life of these patients in the long term. These results were consistent with those of Abouzayd et al. (2016), who found a poor correlation between the outcome of the objective tests and patients’ reported balance symptoms and attributed this to the central compensation for vestibular dysfunction. It would be useful to test balance function over a long postoperative period in order to evaluate the stability of the vestibular compensation for any deficit.

# Conclusions

In this study the sound evoked cVEMP, reflecting saccular function, was most frequently affected by implantation (3 of 7 cases) and was associated with short-term dizziness in 2 patients, although long term vestibular abnormality arising from implantation was not seen. Several patients have abnormal vestibular function pre-implant, so pre-implant multimodal vestibular assessment function may help to inform side of implantation and preserve residual vestibular function post implant. Post-implant assessment including VEMP may help to predict short-term dizziness and target vestibular rehabilitation. This study highlights the potential benefits of conducting multimodal vestibular function pre and post cochlear implantation.

**Conflicts of interest:** None

**References**

Abouzayd, M., Smith, P. F., Moreau, S., & Hitier, M. (2016). What vestibular tests to choose in symptomatic patients after a cochlear implant? A systematic review and meta-analysis. *European Archives of Oto-Rhino-Laryngology*, *274*(1), 53–63.

Alhabib, S. F., & Saliba, I. (2017). Video head impulse test: a review of the literature. *European Archives of Oto-Rhino-Laryngology*, *274*(3), 1215-1222.

Basta, D., Todt, I., Goepel, F., & Ernst, A. (2008). Loss of Saccular Function after Cochlear Implantation: The Diagnostic Impact of Intracochlear Electrically Elicited Vestibular Evoked Myogenic Potentials. *Audiology and Neurotology*, *13*(3), 187–192.

Batuecas-Caletrio, A., Klumpp, M., Santacruz-Ruiz, S., Gonzalez, F. B., Sánchez, E. G., & Arriaga, M. (2015). Vestibular function in cochlear implantation: Correlating objectiveness and subjectiveness. *Laryngoscope*, *125*(10), 2371–2375.

Bell, S. L., Barker, F., Heselton, H., MacKenzie, E., Dewhurst, D., & Sanderson, A. (2015). A study of the relationship between the video head impulse test and air calorics. *European Archives of Oto-Rhino-Laryngology*, 1287–1294.

Berrettini, S., Baggiani, A., Bruschini, L., Cassandro, E., Cuda, D., Filipo, R., ... & Forli, F. (2011). Systematic review of the literature on the clinical effectiveness of the cochlear implant procedure in adult patients. *Acta Otorhinolaryngologica Italica*, *31*(5), 299.

Blakley, B. W., & Wong, V. (2015). Normal Values for Cervical Vestibular-Evoked Myogenic Potentials, 1069–1073.

Curthoys, I. S., & Manzari, L. (2017). Clinical application of the head impulse test of semicircular canal function. Hearing, Balance and Communication, 15(3), 113-126.

Filipo, R., Patrizi, M., La Gamma, R., D’Elia, C., La Rosa, G., & Barbara, M. (2006). Vestibular impairment and cochlear implantation. *Acta Oto-Laryngologica*, *126*(12), 1266–1274.

Forli, F., Arslan, E., Bellelli, S., Burdo, S., Mancini, P., Martini, A., ... & Berrettini, S. (2011). Systematic review of the literature on the clinical effectiveness of the cochlear implant procedure in paediatric patients. *ACTA otorhinolaryngologica italica*, *31*(5), 281.

Fujimoto, C., Egami, N., Kinoshita, M., Sugasawa, K., Yamasoba, T., & Iwasaki, S. (2015). Involvement of vestibular organs in idiopathic sudden hearing loss with vertigo: An analysis using oVEMP and cVEMP testing. *Clinical Neurophysiology*, *126*(5), 1033–1038.

GN Otometrics. (2011). ICS impulse user manual, Doc no. 7-50-1110-EN/01, Denmark.

Handzel, O., Burgess, B. J., & Nadol Jr, J. B. (2006). Histopathology of the peripheral vestibular system after cochlear implantation in the human. *Otology & Neurotology*, *27*(1), 57-64.

Ito, J. (1998). Influence of the multichannel cochlear implant on vestibular function. *Otolaryngology-Head and Neck Surgery*, *118*(6), 900-902.

Jacot, E., Abbeele, T. Van Den, Debre, H. R., Wiener-vacher, S. R., & Pe, H. (2009). Vestibular impairments pre- and post-cochlear implant in children.

Janky, K., & Givens, D. (2015). Vestibular, visual acuity and balance outcomes in children with cochlear implants: a preliminary report. *Ear & Hearing*, *36*(6), e364–e372.

Jin, Y., Shinjo, Y., Akamatsu, Y., Ogata, E., Nakamura, M., Kianoush, S., … Kaga, K. (2008). Vestibular evoked myogenic potentials evoked by multichannel cochlear implant - influence of C levels. *Acta Oto-Laryngologica*, *128*(3), 284–290.

Krause, E., Louza, J. P. R., Wechtenbruch, J., & Gürkov, R. (2010). Influence of cochlear implantation on peripheral vestibular receptor function. *Otolaryngology - Head and Neck Surgery*, *142*(6), 809–813.

Licameli, G., Zhou, G., & Kenna, M. A. (2009). Disturbance of Vestibular Function Attributable to Cochlear Implantation in Children, (April), 740–745.

MacDougall, H. G., Weber, K. P., McGarvie, L. A., Halmagyi, G. M., & Curthoys, I. S. (2009). The video head impulse test Diagnostic accuracy in peripheral vestibulopathy. *Neurology*, *73*(14), 1134-1141.

Maheu, M., Pagé, S., Sharp, A., Delcenserie, A., & Champoux, F. (2017). The impact of vestibular status prior to cochlear implantation on postural control: A multiple case study. *Cochlear Implants International*, *18*(5), 250–255.

Melvin, T. A. N., Della Santina, C. C., Carey, J. P., & Migliaccio, A. A. (2009). The effects of cochlear implantation on vestibular function. *Otology & neurotology: official publication of the American Otological Society, American Neurotology Society [and] European Academy of Otology and Neurotology*, *30*(1), 87.

Migliaccio, A. A., Santina, C. C. Della, Carey, J. P., Niparko, J. K., & Minor, L. B. (2005). The Vestibulo-Ocular Reflex Response to Head Impulses Rarely Decreases after Cochlear Implantation, 655–660.

O'Connell, B. P., Hunter, J. B., & Wanna, G. B. (2016). The importance of electrode location in cochlear implantation. *Laryngoscope investigative otolaryngology*, *1*(6), 169-174.

Psillas, G., Pavlidou, A., Lefkidis, N., Vital, I., Markou, K., Triaridis, S., & Tsalighopoulos, M. (2014). Vestibular evoked myogenic potentials in children after cochlear implantation. *Auris Nasus Larynx*, *41*(5), 432–435.

Robard, L., Hitier, M., Lebas, C., & Moreau, S. (2015). Vestibular function and cochlear implant. *European Archives of Oto-Rhino-Laryngology*, 523–530.

Rosengren, S. M., Welgampola, M. S., & Colebatch, J. G. (2010). Vestibular evoked myogenic potentials: past, present and future. *Clinical Neurophysiology: Official Journal of the International Federation of Clinical Neurophysiology*, *121*(5), 636–51.

Santos, T. G. T., Venosa, A. R., & Sampaio, A. L. L. (2015). Association between Hearing Loss and Vestibular Disorders: A Review of the Interference of Hearing in the Balance. *International Journal of Otolaryngology and Head &amp; Neck Surgery*, *4*(3), 173–179.

Tatovic, M., Babac, S., Djeric, D., Anicic, R., & Ivankovic, Z. (2011). The impact of hearing loss on the quality of life in adults. *Srpski Arhiv Za Celokupno Lekarstvo*, *139*(5-6), 286–290.

Thierry, B., Blanchard, M., Leboulanger, N., Parodi, M., Wiener-Vacher, S. R., Garabedian, E. N., & Loundon, N. (2015). Cochlear implantation and vestibular function in children. *International Journal of Pediatric Otorhinolaryngology*, *79*(2), 101–104.

Tien, H. C., & Linthicum, F. H. (2002). Histopathologic changes in the vestibule after cochlear implantation. *Otolaryngology - Head and Neck Surgery*, *127*(4), 260–264.

Todd, N. P. M., Rosengren, S. M., & Colebatch, J. G. (2008). A source analysis of short-latency vestibular evoked potentials produced by air- and bone-conducted sound. *Clinical Neurophysiology*, *119*, 1881–1894.

Todt, I., Basta, D., & Ernst, A. (2008). Does the surgical approach in cochlear implantation influence the occurrence of postoperative vertigo? 8–12.

Tribukait, A., Brantberg, K., & Bergenius, J. (2004). Function of Semicircular Canals, Utricles and Saccules in Deaf Children. *Acta Oto-Laryngologica*, *124*(1), 41–48.

UK Cochlear Implant Study Group. (2004). Criteria of candidacy for unilateral cochlear implantation is postlingually deafened adults II: Cost-effectiveness analysis. *Ear and Hearing*, *25*(4), 336–360.

Wagner, J. H., Basta, D., Wagner, F., Seidl, R. O., Ernst, A., & Todt, I. (2010). Vestibular and taste disorders after bilateral cochlear implantation. *European Archives of Oto-Rhino-Laryngology: Official Journal of the European Federation of Oto-Rhino-Laryngological Societies (EUFOS): Affiliated with the German Society for Oto-Rhino-Laryngology - Head and Neck Surgery*, *267*(12), 1849–54.

Xu, X.-D., Zhang, X.-T., Zhang, Q., Hu, J., Chen, Y.-F., & Xu, M. (2014). Ocular and cervical vestibular-evoked myogenic potentials in children with cochlear implant. *Clinical Neurophysiology*, *126*(8), 1624–1631.