Vestibular examination in children and adults pre- and post-cochlear implantation

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Masterproef voorgedragen tot het behalen van de graad van master in de logopedische en audiologische wetenschappen
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ABSTRACT - English

Purpose - The first objective of this study was to review the literature to get an overview of the function of the semicircular canals, saccule and utricle before and after cochlear implantation (CI). The second objective of the study was to examine the vestibular function of children and adults before and after CI in order to investigate the influence of CI on vestibular function.

Method – In the literature study, using publications from PubMed and Web of Science about vestibular function before and after CI, results of the rotation test, caloric test, head impulse test, cervical vestibular evoked myogenic potentials (cVEMP) and ocular vestibular evoked myogenic potentials (oVEMP) were integrated. For the prospective study cVEMPs and oVEMPs of six participants between 9 months and 78 years were tested before and after CI.

Results - Results of the literature study showed that 23 to 50% of the cochlear implant patients had preoperative abnormal vestibular test results and that CI caused an increase in the number of patients with vestibular dysfunction. Postoperatively the saccule was most likely to be damaged, followed by the utricle and semicircular canals. Postoperative changes from preoperative normal vestibular function to abnormal function ranged from 0 to 36% of the participants in the literature study and 0 to 50% of the participants in our prospective study, depending on the vestibular test used. Postoperative changes to the saccule and utricle could not be objectified with statistic significant results in our prospective study, yet more absent responses were found postoperatively.

Conclusion - Vestibular dysfunction often occurs in participants with profound hearing loss. Cochlear implantation poses an additional risk for damage to the vestibular system. Preoperative and postoperative vestibular assessment is recommended for all cochlear implant patients and more research, using an extensive test battery, in participants with a CI is needed.
**ABSTRACT - Dutch**

**Doel** – Het eerste doel van deze studie was om de literatuur te doorzoeken om een overzicht te krijgen van de functie van de semicirculaire kanalen, sacculus en utriculus voor en na cochleaire implantatie (CI). Het tweede doel van deze studie was om de vestibulaire functie van kinderen en volwassenen te testen voor en na CI om te onderzoeken wat de invloed is van CI op vestibulaire functie.

**Methode** – In de literatuurstudie werd gebruik gemaakt van publicaties van PubMed en Web of Science over vestibulaire functie voor en na CI. Resultaten van de rotatietest, calorische test, head impulse test en cervicaal vestibulair geëvoikeerde myogene potentialen (cVEMP) en oculair vestibulair geëvoikeerde myogene potentialen (oVEMP) werden geïntegreerd. Voor de prospectieve studie werd de cVEMP en oVEMP afgenomen voor en na CI bij zes deelnemers tussen 9 maand en 78 jaar.

**Resultaten** – Resultaten van de literatuurstudie toonden aan dat 23 tot 50% van de deelnemers aan het onderzoek reeds preoperatieve abnormale vestibulaire resultaten hadden en dat CI een bijkomend risico biedt op vestibulaire dysfunctie. Na CI was de sacculus het meest frequent beschadigd, gevolgd door de utriculus en semicirculaire kanalen. Verandering van preoperatieve normale vestibulaire functie naar abnormale functie trad op bij 0 tot 36% van de deelnemers in de literatuurstudie en bij 0 tot 50% van de deelnemers in onze prospectieve studie, afhankelijk van de vestibulaire test die gebruikt werd. Postoperatieve veranderingen aan de sacculus en utriculus konden niet worden geobjectvieerd met statistisch significante resultaten in onze prospectieve studie, wel werden postoperatief meer afwezige responsen gezien.

**Conclusie** – Vestibulaire dysfunctie treedt vaak op bij personen met een ernstig gehoorverlies. Cochleaire implantaatie zorgt voor een bijkomend risico op schade aan het vestibulair systeem. Preoperatief en postoperatief vestibulair onderzoek is dus aangeraden voor alle patiënten met een CI en meer onderzoek bij personen met een CI, met gebruik van een uitgebreide testbatterij, is noodzakelijk.
1. INTRODUCTION

Since the introduction of cochlear implants (CI) more than 30 years ago, the ability to perceive sound and speech for patients with severe to profound sensorineural hearing loss, can be restored. A cochlear implant is an electronic device that bypasses the damaged auditory sensory hair cells via an electrode array that directly stimulates the auditory nerve fibers (Vincenti et al., 2014). Over the past years, changes have occurred in candidacy for cochlear implantation, such as decreasing age of the candidate, use in patients with residual hearing, growing interest in bilateral implantation and implanting patients with major cochlear malformations and additional handicaps. These changes, in combination with improving satisfactory surgical techniques, have encouraged the use of cochlear implantation (Cohen, 2004). Therefore it is important to consider the possible risks of this procedure.

Because of the close anatomical and phylogenetical relation between the cochlea and the vestibular end-organs, it is likely that sensorineural hearing loss goes hand in hand with abnormalities in vestibular function, even before cochlear implantation (Bonucci, Costa Filho, Mariotto, Amantini, & Alvarenga Kde, 2008; Brey et al., 1995; De Kegel, Maes, Van Waelvelde, & Dhooge, 2015; Tribukait, Brantberg, & Bergenius, 2004). In children with sensorineural hearing loss, the literature indicates that 20-85% demonstrate some type of vestibular dysfunction (Maes, De Kegel, Van Waelvelde, & Dhooge, 2014). The study of Tribukait et al. (2004) studied vestibular function in deaf children. Abnormal function of the horizontal semicircular canals was found in 55% of all participants and saccular and utricular dysfunction in 42% of the participants. In the study of (Zhou, Kenna, Stevens, & Licameli, 2009) 91% of all participants showed abnormal results for vestibular evoked myogenic potentials (VEMP). Consequently, vestibular function should be examined before the implantation to determine preoperative damage to the vestibular system and to determine the risk for additional postoperative damage to the vestibular system. Moreover, results of vestibular testing before implantation can help in the decision of unilateral versus bilateral implantation or the side to be implanted (De Kegel et al., 2015).

It is known that cochlear implantation poses a risk for additional vestibular damage. Vertigo and imbalance are frequently reported complications after cochlear implant surgery (Steenerson, Cronin, & Gary, 2001). In the literature postoperative vestibular
Symptoms occurred in a range of 35% to 74% of the participants (Brey et al., 1995; Filipo et al., 2006; Fina, Skinner, Goebel, Piccirillo, & Neely, 2003; Ito, 1998; Krause, Louza, Wechterbruch, & Gurkov, 2010; Steenerson et al., 2001). Advanced age is found to be a significant factor for developing postoperative vestibular impairment (Brey et al., 1995; Fina et al., 2003; Krause, Louza, et al., 2009; Krause et al., 2010). After cochlear implantation the saccule is most likely to be damaged because of its location close to the pathway of the inserted electrodes, followed by the utricle and semicircular canals (Tien & Linthicum, 2002). In some cases an improvement of vestibular symptoms and function is seen, sometimes in the contralateral ear too, induced by vestibular compensation and electrical stimulation of the vestibular nerve. (Bonucci et al., 2008; Buchman et al., 2004; Ribari, Kustel, Szirmai, & Repasy, 1999). In most of the cases, dizziness is transient (Katsiari et al., 2013; Krause et al., 2010). Nevertheless, these findings indicate that postoperative vestibular changes frequently occur, and should be examined thoroughly.

The objective of this study is to search the literature for abnormal vestibular test results caused by cochlear implantation and to set up the most sensitive test protocol to detect these abnormalities. Secondly, this study will investigate the influence of cochlear implantation on the vestibular function in children and adults by comparing the results of cervical and ocular vestibular evoked myogenic potentials (cVEMP and oVEMP) before and after the implantation. To study the effect of electrical stimulation caused by the cochlear implant, results of VEMPs with the implant switched on and off will be compared.
2. MATERIAL AND METHODS

2.1 Part 1: literature study

The first objective of this study was to search the literature for publications about vestibular function before and after cochlear implantation and to examine which recording parameters are most sensitive to detect vestibular impairment. The purpose of this literature study was to get an overview of function of the semicircular canals as well as both otolith organs before and after cochlear implantation by integrating the test results of the rotation test, caloric test, head impulse test, cervical vestibular evoked myogenic potentials and ocular vestibular evoked myogenic potentials.

2.1.1 Method

A varied search strategy was employed, searching several electronic databases to identify relevant research articles from the peer-reviewed literature. To merit inclusion, relevant full-text articles in English had to relate to vestibular findings before and after cochlear implantation and a clear comparison of pre- and postoperative vestibular test results had to be available.

All relevant articles published before May 2016 were included. Table 1 gives an overview of the databases and search strategies that were used. By using a multifaceted approach, covering the databases PubMed and Web of Science and by cross-referencing with the reference lists in articles, comprehensive coverage was ensured.

After all duplicates had been excluded, remaining articles were screened for eligibility for this literature review.
Table 1. Database and strategy details

<table>
<thead>
<tr>
<th>Database</th>
<th>Search terms</th>
<th>Articles</th>
</tr>
</thead>
<tbody>
<tr>
<td>Web of Science</td>
<td>Cochlear implant AND caloric test</td>
<td>n = 42</td>
</tr>
<tr>
<td>Web of Science</td>
<td>Cochlear implant AND vestibular evoked myogenic potential</td>
<td>n = 37</td>
</tr>
<tr>
<td>Web of Science</td>
<td>Cochlear implant AND head impulse test</td>
<td>n = 10</td>
</tr>
<tr>
<td>Web of Science</td>
<td>Cochlear implant AND rotatory chair</td>
<td>n = 1</td>
</tr>
<tr>
<td>Web of Science</td>
<td>Cochlear implant AND rotation test</td>
<td>n = 1</td>
</tr>
<tr>
<td>PubMed</td>
<td>Cochlear implants (MeSH) OR cochlear implantation (MeSH) AND caloric tests (MeSH)</td>
<td>n = 34</td>
</tr>
<tr>
<td>PubMed</td>
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<td>n = 2</td>
</tr>
<tr>
<td>PubMed</td>
<td>Cochlear implants (MeSH) OR cochlear implantation (MeSh) AND vestibular evoked myogenic potentials (MeSH)</td>
<td>n = 15</td>
</tr>
<tr>
<td>PubMed</td>
<td>Cochlear implants (MeSh) OR cochlear implantation (MeSh) AND rotation test (all fields)</td>
<td>n = 18</td>
</tr>
</tbody>
</table>

Total number of records n = 176

MeSH: Medical Subject Heading

2.2 Part 2: vestibular testing

The second objective of the study was to investigate the influence of cochlear implantation on the vestibular function in children and adults by comparing the results of vestibular tests before and after the implantation.

2.2.1 Participants

For this prospective study participants were recruited from the University of Pretoria Cochlear Implant Unit. These participants all received a cochlear implant in the period between February and May 2016 or already underwent vestibular assessment (cVEMP and oVEMP) before CI and were assessed postoperatively in this period. Information about the participants can be found in table 2. Participant 1 already was implanted on the right side and received during this research an implant on the left side. This study was approved by the Ethical Committee of the University Hospital in Ghent and the Ethical Committee of the University of Pretoria. Informed consents were obtained from all participants or their parents.
2.2.2 Protocol

The protocol for this study was the standard test protocol used at the department of Speech-Language Pathology and Audiology of the University of Pretoria with optimization of the test parameters based on the results of Part 1. For this prospective study vestibular evoked myogenic potentials (VEMPs) were measured.

Before testing, otoscopy and tympanometry were performed to rule out any middle ear problems. Tympanometric results were considered normal if a type A Liden-Jerger classification tympanogram (Jerger, 1970; Liden, 1969) was present. A type A tympanogram is characterized by peak pressure between -100 and +50 daPa, an ear canal volume (ECV) from 0,65 to 1,75 ml for adults and 0,3 to 1 ml for children and a compliance between 0,3 and 1,7 ml in adults and between 0,25 and 1,7 ml in children (Alencar, Iorio, & Morales, 2005).

**Cervical vestibular evoked myogenic potential (cVEMP)**

Cervical vestibular evoked myogenic potential (cVEMP) testing is used to assess saccular function and inferior vestibular nerve function (Xu et al., 2015). This test was performed with the cochlear implant switched on and off. Responses were recorded by surface electrodes (Ambu® BlueSensor N) using an auditory evoked potential system (Bio-logic Navigator Pro, Natus Medical Inc., San Carlos, California, U.S.A.). A non-inverting electrode was placed in the middle of the sternocleidomastoid muscle (SCMm), the inverting electrode at the insertion point of the SCMm to the sternum and a ground electrode on the forehead. The electrode impedance was maintained below 5 kΩ.

<table>
<thead>
<tr>
<th>Number</th>
<th>Age (years)</th>
<th>Sex</th>
<th>Im planted side</th>
<th>Tested side</th>
<th>Test</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Pre CI</td>
<td>Post CI</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1</td>
<td>3,7</td>
<td>3,8</td>
<td>Female</td>
<td>Left</td>
<td>Bilateral cVEMP, oVEMP</td>
</tr>
<tr>
<td>2</td>
<td>0,8</td>
<td>0,9</td>
<td>Male</td>
<td>Bilateral</td>
<td>Left cVEMP</td>
</tr>
<tr>
<td>3</td>
<td>3,2</td>
<td>3,3</td>
<td>Male</td>
<td>Right</td>
<td>Bilateral cVEMP</td>
</tr>
<tr>
<td>4</td>
<td>78,8</td>
<td>79,6</td>
<td>Male</td>
<td>Left</td>
<td>Bilateral cVEMP, oVEMP</td>
</tr>
<tr>
<td>5</td>
<td>3</td>
<td>4,1</td>
<td>Female</td>
<td>Bilateral</td>
<td>Bilateral cVEMP</td>
</tr>
<tr>
<td>6</td>
<td>49,1</td>
<td>50,3</td>
<td>Male</td>
<td>Left</td>
<td>Bilateral cVEMP, oVEMP</td>
</tr>
</tbody>
</table>
Adults were tested in supine position with the head turned to the contralateral side while raising the head. Infants and children were tested in sitting position with their heads rotated as far as possible to the side contralateral to the stimulated ear. Contraction was maintained by attracting the child’s attention with toys, audiovisual stimulation or parents calling their infant from the opposite direction. Those maneuvers activated the ipsilateral SCM muscle (Sheykholeslami, Megerian, Arnold, & Kaga, 2005). Constant muscle tension (150 µV to 250 µV) is important (Maes et al., 2014).

Alternating linear 500 Hz tone bursts with a rise/fall time of 1 ms and 2 ms plateau were presented monaurally through insert earphones at an intensity level of 95 dB nHL (130 dB SPL) or through a bone conductor at an intensity of 60 dB nHL with a stimulus rate of 5.1/s. The band-pass intensity was 10-1500 Hz and the electromyographic signal was amplified 5000 times. Two trials were recorded of each cVEMP response consisting of 100 averaged sweeps.

A biphasic short-latency wave with a positive peak P1 (p13) and negative peak N1 (p23) was considered as a normal VEMP tracing. We analyzed the absolute latencies of P1 and N1 as the time (ms) between the maximal peaks of P1 and N1 and t=0ms. The amplitude of P1 and N1 and interpeak amplitude (µV) was determined and the asymmetry ratio (%) was calculated using the interaural peak to peak differences divided by the total peak to peak amplitude of both ears.

**Ocular vestibular evoked myogenic potential (oVEMP)**

Ocular vestibular evoked myogenic potential (oVEMP) testing is used to assess utricular function and superior vestibular nerve function (Xu et al., 2015). This test was performed with the cochlear implant switched on and off. Responses were recorded by surface electrodes (Ambu® BlueSensor N) using the same auditory evoked potential system as for cVEMP testing. A non-inverting electrode was placed under the eye on the inferior oblique, the inverting electrode on the side of the nose bridge and a ground electrode on the forehead. The electrode impedance was maintained below 5 kΩ.
All participants (including children) were tested in supine position and were asked to gaze upwards with a vertical visual angle of approximately 30° above horizontal to a marked point upon presentation of the sound stimulus. In small children attention-getting toys were used to obtain the correct eye position. The studies of Wang, Hsieh, and Young (2013) and Young (2015) showed that the otolithic-ocular reflex is not yet matured before the age of two years and consequently repeatable oVEMP responses cannot be elicited. Since this reflex is necessary for the development of independent gait in children, the oVEMP test in our study was only used when the child could walk independently and was older than two years to make sure the otolithic-ocular reflex was matured.

Alternating linear 500 Hz tone bursts with a rise/fall time of 1 ms and 2 ms plateau were presented monaurally through insert earphones at an intensity level of 95 dB nHL (130 dB SPL) or through a bone conductor at an intensity of 60 dB nHL with a stimulus rate of 5.1/s. The band-pass intensity was 1-500 Hz, the electromyographic signal was amplified 100 000 times. Two trials were recorded of each oVEMP response consisting of 100 averaged sweeps.

A biphasic short-latency wave with a negative peak N1 (n10) and positive peak P1 (p15) was considered as a normal oVEMP tracing. We analyzed the absolute latencies of N1 and P1 latencies as the time (ms) between the maximal peaks of N1 and P1 and t=0ms. The amplitude of P1 and N1 and interpeak amplitude (µV) was determined and the asymmetry ratio (%) was calculated using the interaural peak to peak differences divided by the total peak to peak amplitude of both ears.
**Data analysis**

Statistical analysis was performed using IBM SPSS Statistics 23 (SPSS Inc., Chicago IL). The non-parametric Related Samples Wilcoxon Signed Rank Test was used to identify if there was a significant difference (p < 0.05) between results for the parameters latency of P1, latency of N1, amplitude of P1, amplitude of N1, interpeak amplitude of oVEMP and cVEMP testing in three different conditions. First a comparison was made for each of these parameters between the pre- and postoperative results of the implanted side with the implant switched off. Secondly this statistical test was used to compare the pre- and postoperative results on the implanted side with the implant switched on. Furthermore, the Wilcoxon test was used to identify a difference in latency of P1, latency of N1, amplitude of P1, amplitude of N1, interpeak amplitude in the postoperative condition with the implant switched on and off. The Wilcoxon test was also used for comparison of the pre- and postoperative results on the non-implanted side. The same statistical test was used to compare the asymmetry ratio in the preoperative and postoperative condition with the implant switched on and off and to compare both postoperative conditions.
3. RESULTS

Figure 1 summarizes the different phases to identify relevant articles for this literature review. Implementation of different search strategies in the databases Web of Science and PubMed resulted in identification of 176 records in total. After removal of duplicates, 98 articles were screened for relevance by reading titles and abstracts. Sixty-nine articles were excluded. Reasons for exclusion of these articles are described in figure 1. The full text of the 29 remaining articles and three articles that were found by cross-referencing with the reference lists in articles, were assessed for eligibility. Thirteen full-text records were excluded with reasons, described in figure 1.

In total, 19 articles were directly relevant for this literature review. In eight of these studies the pre- and postoperative results of the cervical vestibular evoked myogenic potential (cVEMP) test were described for 60 children and 95 adults. Only one study (Xu et al., 2015), including 31 children, used the ocular vestibular evoked myogenic potential (oVEMP) test in his protocol. The results of the caloric test before and after CI were determined in 12 studies, taking into account 281 participants. In two of these studies, which included 47 participants in total, no distinction was made between children and adults when describing the results. The remaining 234 participants were all adults. The effect of CI on the bedside head impulse test was only described in one study (Thierry et al., 2015), including 12 children. In five studies, including 98 participants in total, results of the rotation test were available. No distinction was made between children and adults when describing the results.
Records identified through database Web of Science (n = 107)

Records identified through database PubMed (n = 69)

Records identified through databases (n = 176)

Records after duplicates removed (n = 98)

Records screened (title and abstract) (n = 98)

Records excluded, with reasons:
- Animal studies (n = 2)
- No vestibular test results (VEMP, HIT, caloric test or rotation test) (n = 13)
- Not about cochlear implantation (n = 15)
- Only preoperative results (n = 10)
- Only postoperative results (n = 8)
- No full text (n = 11)
- Not in English (n = 1)
- Cochlear implant failure (n = 2)
- Case report (n = 2)
- Electric acoustic stimulation (EAS) cochlear implantation (n = 1)
- Focus on insertion depth (n = 3)
- Focus on inner ear malformation (n = 1)

(n = 69)

Records obtained from references (n = 3)

Full-text articles assessed for eligibility (n = 32)

Full-text records excluded, with reasons:
- Test results not described separately for each vestibular test (n = 3)
- Unclear postoperative results (n = 1)
- Only postoperative results when preoperative results were normal (n = 1)
- No comparison of preoperative and postoperative results per participant (n = 8)

(n = 13)

Studies included in review (n = 19)

Figure 1. Phases for identification of relevant reports for inclusion
3.1 Cervical vestibular evoked myogenic potential (cVEMP)

Table 3 shows the effect of cochlear implantation on the results of the cVEMP test according to the literature. Results of cVEMP testing were considered abnormal from the moment the latency of P1 and N1 was delayed, the amplitude was decreased or a reproducible bilateral response was absent. In seven of the eight studies an increase in abnormal cVEMP results was seen, while in one study (De Kegel et al., 2015) no change occurred. Preoperative testing showed an abnormal result in 55 of 155 participants (35%) included in this table. Preoperative results were comparable between children (33%) and adults (37%). After cochlear implantation, abnormal results were present in 111 of 155 participants (72%). All 55 participants (35%) with preoperative abnormal results also showed abnormal results after cochlear implantation. Fifty-six of 155 participants (36%) showed a decrease from preoperative normal to postoperative abnormal results. In six of the eight studies, amplitude was considered to be the most important parameter. To determine a decrease of the cVEMP response these studies mainly focused on the amplitude and the waveform of the response.

In the literature two studies determined cVEMP responses with the device switched on and off. Eleven of twelve participants in the study of Jin, Nakamura, Shinjo, and Kaga (2006) showed no cVEMP responses with the device switched off and three participants showed a change towards present cVEMP responses with the device switched on. One participant had responses with CI switched on as well as off, although the amplitude was lower with the device off. In the study of Psillas et al. (2014), however, responses remained absent both with CI switched on and off.
Pre-post analysis of the implant ear in our prospective study, showed absent cVEMP responses in one of six participants (17%) (table 4). These responses remained absent after implantation. Three of six participants (50%) showed a change from preoperative present towards postoperative absent cVEMP responses. Measurements of cVEMP responses with the implant switched on and off showed no significant differences.

Since two of the six participants were implanted bilaterally, four non-implant ears could be taken into account. Pre-post analysis of the non-implant ear, showed absent cVEMP responses in one of four participants (25%), which remained absent after implantation. One of four participants (25%) showed a change from preoperatively present to postoperatively absent cVEMP response.

Because of the small sample size, statistical analysis on the different cVEMP response parameters could not be performed.

<table>
<thead>
<tr>
<th>Table 3. Effects of cochlear implantation on cVEMP</th>
</tr>
</thead>
<tbody>
<tr>
<td>Reference</td>
</tr>
<tr>
<td>De Kegel (2015)*</td>
</tr>
<tr>
<td>Jin (2015)*</td>
</tr>
<tr>
<td>Krause (2009)</td>
</tr>
<tr>
<td>Katsiari (2013)</td>
</tr>
<tr>
<td>Licameli (2009)*</td>
</tr>
<tr>
<td>Psillas (2014)*</td>
</tr>
<tr>
<td><strong>Total</strong></td>
</tr>
<tr>
<td>Children*</td>
</tr>
<tr>
<td>Adults</td>
</tr>
</tbody>
</table>

**cVEMP**: cervical vestibular evoked myogenic potential

* Test subjects were children

<table>
<thead>
<tr>
<th>Table 4. Effects of cochlear implantation on cVEMP – implanted side</th>
</tr>
</thead>
<tbody>
<tr>
<td>Reference</td>
</tr>
<tr>
<td>This study</td>
</tr>
</tbody>
</table>
3.2 Ocular vestibular evoked myogenic potential (oVEMP)

The study of Xu et al. (2015) is the only one to report data of oVEMPs after cochlear implantation. In children, the oVEMP responses of the implanted ear with the CI switched on and off were compared. Preoperatively, 6 of 26 children (23%) had abnormal oVEMP responses and after CI 21 of 26 (81%) had abnormal results when the device was switched off. Preoperatively, 9 of 31 children (29%) had abnormal oVEMP response compared to 27 of 31 children (87%) who had abnormal results with the device switched on. It is however not clear how many children with postoperative abnormal results already had preoperative abnormal results. On the implanted side, elevation of thresholds, decrease of amplitudes, prolonged latencies of N1 and P1 and shorter interpeak amplitudes were noticed.

<table>
<thead>
<tr>
<th>Reference</th>
<th>Preoperative abnormal (%)</th>
<th>Postoperative abnormal (%)</th>
<th>Parameter</th>
</tr>
</thead>
<tbody>
<tr>
<td>Xu (2015)*</td>
<td>9 of 31 (29)</td>
<td>27 of 31 (87) - ON</td>
<td>Amplitude, latencies, thresholds, interpeak latencies</td>
</tr>
<tr>
<td></td>
<td>6 of 26 (23)</td>
<td>21 of 26 (81) – OFF</td>
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</tbody>
</table>

oVEMP: ocular vestibular evoked myogenic potential
* Test subjects were children

<table>
<thead>
<tr>
<th>Table 5. Effects of cochlear implantation on oVEMP – implanted side</th>
</tr>
</thead>
<tbody>
<tr>
<td>Reference</td>
</tr>
<tr>
<td>-------------</td>
</tr>
<tr>
<td>This study</td>
</tr>
</tbody>
</table>

In our prospective study ocular vestibular evoked myogenic potentials (oVEMP) could be recorded bilaterally for three of the six participants. In two participants responses were bilaterally present both before and after cochlear implantation. In one of three participants (33%) oVEMP responses were absent both before CI and after implantation. Measurements of oVEMP responses with the implant switched on and off showed no significant different responses.
Results of the non-implant ear of these three participants are as follows. In one participant responses were absent before CI and present afterwards. In two participants oVEMP responses were present both before and after CI.

Because of the small sample size, statistical analysis on the different cVEMP response parameters could not be performed.

### 3.3 Caloric test

In table 7 results of pre- and postoperative results for caloric testing, found in the literature, are presented. All studies used water stimulation rather than air stimulation, except Katsiari et al. (2013). Results were considered abnormal when the slow component velocity (SCV) was abnormally low or when unilateral weakness or directional preponderance was noticed.

In seven of twelve studies, participants with preoperative abnormal results also showed abnormal postoperative results. In four studies (Brey et al., 1995; Filipo et al., 2006; Katsiari et al., 2013; Krause, Louza, et al., 2009), however, one participant with preoperative abnormal results regained normal results after cochlear implantation. In the study of Robard, Hitier, Lebas, and Moreau (2015) even five individuals changed from abnormal results to normal results. In all studies, however, an increase in the total amount of participants with abnormal caloric test results was seen.

Before cochlear implantation, 135 of 281 participants (48%) had abnormal results whereas postoperatively, 190 of 281 participants (67%) showed abnormal results. Of all participants 63 (23%) showed a decrease from preoperative normal to postoperative abnormal results.

In the study of Bonucci et al. (2008) only results of the implanted side were described. Of the 13 individuals (44%) with normal preoperative results, 8 showed postoperative abnormal results. It is not known if there was any change in the results of people with preoperative abnormal results. The article only indicated that 4 individuals with hyporeflexia started having areflexia on the implanted side.
Table 7. Effects of cochlear implantation on caloric test

<table>
<thead>
<tr>
<th>Reference</th>
<th>Preoperative abnormal (%)</th>
<th>Postoperative abnormal (%)</th>
<th>Parameter</th>
</tr>
</thead>
<tbody>
<tr>
<td>Basta (2008)</td>
<td>2 of 18 (11)</td>
<td>3 of 18 (17)</td>
<td>UW</td>
</tr>
<tr>
<td>Bonucci (2008)*</td>
<td>25 of 38 (66)</td>
<td>31 of 38 (81)</td>
<td>SCV</td>
</tr>
<tr>
<td>Brey (1995)</td>
<td>5 of 17 (29)</td>
<td>9 of 17 (53) ~</td>
<td>SCV, UW</td>
</tr>
<tr>
<td>Filipo (2006)</td>
<td>8 of 14 (57)</td>
<td>10 of 14 (71) ~</td>
<td>DP, SCV</td>
</tr>
<tr>
<td>Ito (1998)</td>
<td>18 of 35 (51)</td>
<td>23 of 35 (66)</td>
<td>SCV</td>
</tr>
<tr>
<td>Katsiari (2013)</td>
<td>13 of 20 (65)</td>
<td>15 of 20 (75) ~</td>
<td>SCV, UW</td>
</tr>
<tr>
<td>Krause (2009)</td>
<td>12 of 24 (50)</td>
<td>19 of 24 (79)</td>
<td>SCV</td>
</tr>
<tr>
<td>Krause (2009)</td>
<td>16 of 38 (42)</td>
<td>24 of 38 (63) ~</td>
<td>SCV, UW, DP</td>
</tr>
<tr>
<td>Krause (2010)</td>
<td>14 of 30 (47)</td>
<td>22 of 30 (73)</td>
<td>SCV, UW</td>
</tr>
<tr>
<td>Robard¹ (2015)</td>
<td>13 of 30 (43)</td>
<td>21 of 30 (70)</td>
<td>SCV, UW</td>
</tr>
<tr>
<td>Van Den Broek (1993)</td>
<td>5 of 6 (83)</td>
<td>6 of 6 (100)</td>
<td>SCV</td>
</tr>
<tr>
<td>Vibert (2001)°</td>
<td>4 of 9 (44)</td>
<td>5 of 9 (55)</td>
<td>SCV</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td>135 of 279 (48)</td>
<td>188 of 279 (67)</td>
<td></td>
</tr>
</tbody>
</table>

SCV: slow component velocity; UW: unilateral weakness; DP: directional preponderance
~ One individual with preoperative abnormal results had postoperative normal results
¹ Five individuals with preoperative abnormal results had postoperative normal results
*Test subjects were children and adults

3.4 Head impulse test (HIT)

Table 8 shows the effects of cochlear implantation on the head impulse test. Only the study of Thierry et al. (2015) used bedside head impulse testing in their protocol for vestibular assessment and compared the postoperative to the preoperative findings. Preoperative abnormal results were present in 6 of 12 (50%) participants. After cochlear implantation, the results of five of the participants with abnormal preoperative vestibular function remained abnormal. An improvement of the results was seen in one patient with abnormal results before CI. No participants with preoperative normal results showed a VOR impairment after implantation. To assess the horizontal canal this study observed the presence of corrective saccades.

Table 8. Effects of cochlear implantation on HIT

<table>
<thead>
<tr>
<th>Reference</th>
<th>Preoperative abnormal (%)</th>
<th>Postoperative abnormal (%)</th>
<th>Parameter</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Preoperative normal</td>
<td>Preoperative abnormal</td>
<td></td>
</tr>
<tr>
<td>Thierry (2015)*</td>
<td>6 of 12 (50)</td>
<td>0 of 12 (0)</td>
<td>5 of 12 (42)</td>
</tr>
</tbody>
</table>

HIT: head impulse test
*Test subjects were children
3.5 Rotation test

The results of rotational testing, in participants included in the literature study, before and after CI are shown in table 9. Results were considered abnormal when a low gain or directional preponderance was noticed. The results of all individuals with abnormal function before implantation remained abnormal, except for one individual in the study of Brey et al. (1995) and two individuals in the study of Krause et al. (2009) who showed a postoperative change towards normal results.

Preoperatively, 47 of 98 participants (48%) had abnormal results and postoperatively 60 of 98 participants (61%) had abnormal test results. Postoperatively, 16 of all 98 participants (16%) showed a change from normal preoperative results towards abnormal results on the rotation test.

<table>
<thead>
<tr>
<th>Reference</th>
<th>Preoperative abnormal (%)</th>
<th>Postoperative abnormal (%)</th>
<th>Parameter</th>
</tr>
</thead>
<tbody>
<tr>
<td>Abramides, Bittar, Tsuji, and Bento (2015)°</td>
<td>17 of 21 (81)</td>
<td>17 of 21 (81)</td>
<td>NI</td>
</tr>
<tr>
<td>Brey (1995)</td>
<td>8 of 13 (61)</td>
<td>8 of 13 (61) ~</td>
<td>Gain, phase</td>
</tr>
<tr>
<td>Filipo (2006)*</td>
<td>2 of 17 (12)</td>
<td>3 of 17 (18)</td>
<td>DP</td>
</tr>
<tr>
<td>Krause (2009)¹</td>
<td>16 of 36 (44)</td>
<td>25 of 36 (70)</td>
<td>DP</td>
</tr>
<tr>
<td>Vibert, Hausler, Kompis, and Vischer (2001)°</td>
<td>4 of 11 (36)</td>
<td>7 of 11 (64)</td>
<td>NI</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td><strong>47 of 98 (48)</strong></td>
<td><strong>60 of 98 (61)</strong></td>
<td></td>
</tr>
</tbody>
</table>

DP: directional preponderance; NI: not indicated
~ One individual with preoperative abnormal results had postoperative normal results
¹ Two individuals with preoperative abnormal results had postoperative normal results
°Test subjects were children and adults
* Before activation cochlear implant
4. DISCUSSION

The cochlea and vestibule, which are located close to each other, share a continuous membranous labyrinth structure. Furthermore both systems have similar receptor cells and receive blood supply from a common arteria (Tribukait et al., 2004; Xu et al., 2016; Xu et al., 2015). Therefore it is likely that damage occurs to both structures simultaneously. The first objective of this study was to review the literature to get an overview of the function of the semicircular canals, saccule and utricle before and after cochlear implantation (CI). The second objective of the study was to test vestibular function of children and adults before and after CI in order to investigate the influence of CI on vestibular function.

Preoperative vestibular function

It is known from the literature that participants with profound sensorineural hearing loss often show vestibular impairments (Bonucci et al., 2008; Brey et al., 1995; De Kegel et al., 2015; Tribukait et al., 2004). None of the studies, however, that were included in this literature study assessed vestibular function using rotatory, caloric, HIT, cVEMP as well as oVEMP results all together, before and after CI. Only a selection of these tests was used. The purpose of this literature study was to integrate the test results of all these separate studies.

In the literature study cVEMP, oVEMP, caloric, HIT as well as rotatory results before and after implantation were reported for each study and vestibular test separately (Table 3, 5, 7, 8, 9) and showed a wide variety in the amount of participants who had preoperative vestibular dysfunction (11% to 83%). Reasons for diversity of these results are most likely the result of differences in the used test battery.

In our literature study it was noticed that the horizontal semicircular canals are most likely to show a deficit before cochlear implantation. The head impulse test, caloric test and rotation test all assess functioning of the horizontal semicircular canals and showed abnormal results in respectively 50%, 48% and 48% of all participants. Horizontal canal dysfunction was followed by saccular dysfunction in 35% of the participants who underwent cVEMP testing. These results were supported by the study of Tribukait et al. (2004) where horizontal semicircular canal function, as well as functioning of the utricle and saccule in deaf children was assessed. Caloric testing
showed abnormal functioning of the semicircular canals in 45% of all children whereas saccular and utricular dysfunction was present in 42% of participants. In the study of Xu (2016) oVEMP, cVEMP and caloric test results of adults who were diagnosed with congenital bilateral profound sensorineural hearing loss were available. Forty ears (74%) showed abnormal results for caloric testing. Utricular damage could be observed in 61% and saccular dysfunction in 55%. Results of oVEMP testing in this literature study showed a preoperative abnormal function in 29% of the participants. This result, however, was only based on one study with a limited amount of participants. In our prospective study responses for cVEMP and oVEMP testing were preoperatively absent in 17 to 33% of the participants. These numbers are rather low in comparison to other studies but this could possibly be the result of the small amount of participants in our prospective study.

Postoperative vestibular findings

Cochlear implantation enhances the risk for vestibular dysfunction because insertion of the electrodes in the scala tympani may damage the spiral ligament and stria vascularis or the basilar membrane, increasing the risk of endolymph mixing with perilymph (Filipo et al., 2006; Huygen et al., 1995). Other possible histopathologic changes in consequence of surgery are fibrosis, reactive neuromas (Tien & Linthicum, 2002), new bone formation (Nadol et al., 2001) and acute transient labyrinthitis (Fina et al., 2003). A lot of studies about influence of cochlea implantation on the balance system, however, use different test groups in their research in terms of etiology of the hearing loss, age of participants and amount of participants included.

Vestibular symptoms often occur after cochlear implantation. In the literature postoperative vestibular symptoms occurred in a range of 35% to 74% of the participants (Brey et al., 1995; Filipo et al., 2006; Fina et al., 2003; Ito, 1998; Krause et al., 2010; Steenerson et al., 2001). According to the study of Tien et al. (2002), the saccule is most frequently damaged after cochlear implantation because of its close location to the insertion place of the electrodes, followed by the utricle and semicircular canals. The results of our literature study were consistent with that. We found that 36% of the participants with preoperative normal functioning of the saccule showed abnormal postoperative test results for cVEMP testing. In our prospective study 50% of all participants with preoperative normal saccule function had abnormal results.
postoperatively. One of the participants with both preoperative and postoperative an absent cVEMP response was already 78 years old at the first test moment. Since cVEMP responses decrease around the age of 60 (Janky & Shepard, 2009), it is not possible to determine if the absent response was the result of age or vestibular dysfunction correlated with the hearing loss.

In the literature two studies determined the cVEMP response with the device switched on and off. Eleven of twelve participants in the study of Jin et al. (2006) showed no cVEMP responses with the device switched off and three participants showed a change towards cVEMP responses with the device switched on. One participant had responses both with CI switched on and off, although the amplitude was lower with the device off. These findings may be explained by vestibular compensation and electrical stimulation of the vestibular nerve (Bonucci et al., 2008; Buchman et al., 2004; Ribari et al., 1999). In the study of Psillas et al. (2014), however, responses remained absent both with CI switched on and off. In the study of Xu et al. (2015) a higher rate of vestibular dysfunction was found with the device switched on for oVEMP testing. Utricular dysfunction was present after implantation in 87% of all participants when the CI was switched on. With the cochlear implant switched off 81% showed abnormal utricular function. These results, however, are only based on one research study and therefore cannot be objectively taken into account. Furthermore it was not clear if the participants with postoperative utricular dysfunction also had abnormal results preoperatively so we cannot conclude what the effect of CI on oVEMP is. In our prospective study no change occurred after cochlear implantation. Both before and after CI only one of the participants showed no response. Only three participants were included for oVEMP testing in this study so this is not enough to draw any conclusions. This emphasizes that further research about oVEMPs in people who use a cochlear implant is needed.

The hypothesis that after CI latencies are prolonged and (interpeak) amplitudes on the implanted side are decreased, could not be objectified during this prospective study. No statistical significant differences were found when comparing the latency of P1, latency of N1, amplitude of P1, amplitude of N1, interpeak amplitude and asymmetry ratio for the three different conditions (preoperative and postoperative with implant off/on and postoperative with implant off and on) both for cVEMP and oVEMP.
In the literature study, the number of participants with postoperative abnormal function of the horizontal semicircular canals is lower than participants with damage to the utricle or saccule even though. For the caloric test and rotation test 23% and 16% of all participants with preoperative normal results showed a vestibular dysfunction after cochlear implantation.

Only one research study used the bedside head impulse test (HIT) to assess pre- and postoperative vestibular function. In this study only 12 children were included. VOR-impairment did not increase after cochlear implantation and one participant even showed an improvement. It is, however, possible that more participants had a VOR impairment because covert saccades could be missed when using the bedside head impulse test. The fact that less participants show a change in function of the horizontal canal could, however, also be explained by the large amount of participants who already had abnormal function preoperatively.

Limitations of study and suggestions for further research

This study has a few limitations. First of all only a limited amount of participants was included in our prospective study, six participants for cVEMP and three for oVEMP. Furthermore, the test group was not uniformly distributed and included both adults and children. Therefore it is not possible to draw conclusions from these test results. It is recommended that further research is done using much more participants.

This literature study showed that vestibular testing before and after cochlear implantation often is limited in the amount of tests used. In this prospective study only cVEMP and oVEMP were used. It is clear that a more extensive test battery is needed in the evaluation of preoperative and postoperative vestibular function. It is recommended that each part of the vestibular system (saccule, utricle and semicircular canals) is assessed separately. This will give a better overview of vestibular function before and after CI because in that way, all parts of the vestibular system are evaluated. In adults vestibular assessment with the use of at least caloric testing, cVEMP and oVEMP is recommended. If available, video head impulse testing can be performed because this gives information about higher frequencies of the VOR (Hülse, Hörmann, Servais, Hülse & Wenzel, 2015). For children, however, caloric testing is often not possible because of a lack of cooperation and fear. To assess horizontal
canal function in children the rotation test is recommended. Utricular and saccular function in children can be assessed using cVEMP and oVEMP testing.

Furthermore the etiology of hearing loss was not included in this study. In further research with a large test group it could be useful to document the etiology of hearing loss in order to link this to preoperative vestibular dysfunction.
5. Conclusion

Results of the literature as well as the prospective study both show that preoperative vestibular dysfunction often occurs in candidates for CI (23-50%) and that cochlear implantation poses a risk for more vestibular damage. Before CI the horizontal semicircular canals are most likely to show a deficit, followed by the saccule and utricle. After implantation the saccule is most frequently damaged followed by the utricle and semicircular canals. Postoperative changes from preoperative normal vestibular function to abnormal function ranged from 0 to 36% of the participants in the literature study and 0 to 50% of the participants in our prospective study, depending on the vestibular test.

Therefore, it is important that paediatric and adult CI candidates are assessed before and after implantation using an extensive test battery. Especially in children this is important because it is known that hearing impaired children may have a delay in gross motor development, which may be more severe if a vestibular problem is superimposed to the auditory problem. This way, early motor intervention can be started if necessary and parents can be better prepared for possible consequences of cochlear implantation. Also for adults, it is important to gain insight into their pre- and postoperative vestibular test results, in order to start appropriate rehabilitation programs to improve their quality of life. Preoperative results of vestibular function in both ears can also help in the decision-making about which ear to implant in case of unilateral CI in order to preserve as much vestibular function as possible. All of this emphasizes the fact that more research is needed on a large patient population with an extensive vestibular test battery and a comparison between test results with the CI switched on and off should be made in order to identify the impact of a functioning CI on the vestibular function.
6. References


