Characterization of the vestibular function in an otosclerosis population – a pilot case-control study

Original Article

Authors

Miguel Campos Lopes
Centro Hospitalar de Entre o Douro e Vouga, Portugal

Alexandra Correia
Centro Hospitalar de Entre o Douro e Vouga, Portuga

Hélder Silva
Centro Hospitalar de Entre o Douro e Vouga, Portugal

Natália Oliveira
Centro Hospitalar de Entre o Douro e Vouga, Portugal

Diana Ribeiro
Centro Hospitalar de Entre o Douro e Vouga, Portugal

Vanessa Neves
Centro Hospitalar de Entre o Douro e Vouga, Portugal

Vera Miranda
Centro Hospitalar de Entre o Douro e Vouga, Portugal

Carlos Carvalho
Centro Hospitalar de Entre o Douro e Vouga, Portugal

Correspondence:
Miguel Campos Lopes
miguelcamposlopes@outlook.com

Article received on April 9, 2023.
Accepted for publication on May 26, 2023.

Abstract

Objectives: Characterization of the vestibular function in an otosclerosis population, and its comparison with a healthy population.
Study Design: Case-control study.
Material and Methods: Patients with clinical/audiological suspicion of otosclerosis (surgically confirmed) and healthy controls matched for age and sex were recruited. Vestibular function was evaluated using the Video Head Impulse Test and the Dizziness Handicap Inventory (DHI) questionnaire.
Results: A total of 24 participants (12 cases and 12 controls) were included, of whom 91.6% were female, with a mean age of 44±2 years. The mean semicircular canal gain for each canal did not show a statistically significant difference between the two groups. On the other hand, the DHI scores differed significantly between the two groups (24.1 vs. 6.3; Mann-Whitney $p<0.01$).
Conclusion: It is possible to verify a subjective functional compromise of balance in patients with otosclerosis, which does not appear to be associated with dysfunction of the semicircular canals.

Keywords: Otosclerosis; Vestibular Function; Video Head Impulse Test; Dizziness Handicap Inventory.

Introduction

Otosclerosis is one of the most common otologic diseases, with an estimated prevalence of 0.3–0.4%. It affects the female sex predominantly and typically presents in the third and fourth decades of life. Its clinical course is characterized by progressive conductive hearing loss secondary to otosclerotic footplate fixation, which can progress to cochlear involvement and sensorineural hearing loss.[1]

The hearing component of otosclerosis is well known and comprehensively described in the literature.[2,3] However, the evidence regarding

vestibular involvement is scarce. Recent studies have suggested that there may be some degree of involvement of the vestibular system in patients with otosclerosis, which may be detected through a comprehensive vestibular evaluation—through the video head impulse test (vHIT), caloric tests, and cervical and ocular vestibular evoked myogenic potentials (cVEMPs and oVEMPs).[4–6] Nevertheless, there is no consensus on the etiology and pathophysiology of vestibular involvement or the best method to assess it.

This study aimed to characterize the vestibular function in patients with otosclerosis who had not been operated on, using the vHIT and quality of life questionnaire related to vestibular symptoms using the Dizziness Handicap Inventory (DHI) and compare this sample with a healthy population.

Materials and Methods

Sample

This was a cross-sectional observational case-control study. The participants were recruited between October 2022 and February 2023 and divided into two groups. The first group (cases) comprised patients with otosclerosis referred from the otorhinolaryngology clinic of a tertiary hospital (Centro Hospitalar de Entre o Douro e Vouga) who were recruited using consecutive sampling.

The inclusion criteria were as follows: clinical and audiological suspicion of otosclerosis (evidence of conductive hearing loss with an intact tympanic membrane); unilateral or bilateral disease in unoperated ears; diagnosis confirmed intraoperatively a posteriori. The exclusion criteria were as follows: presence of vestibular disease; previous history of otologic surgery; chronic otitis media; absence of surgical confirmation of the diagnosis.

The second group comprised controls—healthy patients recruited at the hospital through convenience sampling—who had no previous history of otologic or vestibular disease; they were matched for age and sex.

Vestibular evaluation

Vestibular function was assessed preoperatively using the vHIT system (ICS Impulse System, GN Otometrics). After the initial evaluation for the presence of spontaneous nystagmus, the device was calibrated. The examination was performed with the acquisition of 20 head impulses in each direction in each of the three vestibular planes—horizontal, RALP (right anterior/left posterior), and LARP (left anterior/right posterior). The criteria used to establish the dysfunction of the semicircular canal were gains lower than 0.8 (in the horizontal semicircular canals) or 0.7 (in the vertical semicircular canals), asymmetry greater than 15%, and the presence of corrective saccades (overt and covert), defined as pathological according to the system's software, after visual inspection of the graph to exclude artifacts.[7] The examinations were performed by a trained physician or audiologist.

Additionally, the DHI questionnaire—the version that has been translated and validated in Portuguese[8]—was administered. It comprises 25 items (scored from 0 to 4) on the subjective evaluation of vestibular and balance symptoms and, consequently, the quality of life associated with these symptoms. This questionnaire is subdivided into three domains: physical, functional, and emotional.

Statistical analysis

Descriptive analysis was used to document the characteristics of the study sample. Normal distribution was assessed through visual analysis of the histogram. Categorical variables are expressed as frequencies and percentages (n, %), and normal continuous variables are expressed as means ± standard deviation (SD). The non-parametric Mann-Whitney, Wilcoxon, and Kruskal-Wallis tests were used to compare the mean gain of the vestibulo-ocular reflex (VOR) of the semicircular canals between the groups, and the Mann-Whitney test was used to compare the scores obtained in the DHI. Fisher's exact test was used to compare the presence of corrective saccades in the vHIT between the groups.

All data were processed and analyzed using the IBM SPSS Statistics software , version 27, and statistical significance was set at p<0.05.

Informed consent

All procedures in this study were performed according to the regulations established by the institution's Clinical Research and Ethics Committee and the Declaration of Helsinki of 1964 of the World Medical Association and its later revisions. The study received a positive opinion and approval from the aforementioned ethics committee, with CES no. 57_2022. All participants signed the informed consent form for the study.

Results

Sample characterization

The final sample contained 24 participants—12 each in the otosclerosis and control groups. Twenty-two participants (91.6%) were women, and the mean age of the two groups was 44 ± 8 years. Six patients (50%) in the otosclerosis group were under clinical and audiological suspicion of having bilateral disease at the time of diagnosis, and none showed evidence of cochlear otosclerosis[9] (defined as sensorineural or mixed hearing loss attributable to the disease).

Table 1 shows the clinical and demographic characteristics of the sample.

Comparative analysis of vHIT results

The mean VOR gains of the horizontal semicircular canals in the otosclerosis and control groups were 0.97 and 1.04 for the right ear and 0.90 and 0.97 for the left ear, respectively. This difference was not statistically significant (Man-Whitney p=0.89 on the right and p=0.76 on the left). The results obtained for the other semicircular canals and presence of corrective saccades were also not statistically significant (Fisher p=0.32). These results are shown in Table 2.

Subsequently, subgroup analysis was performed to compare the VOR gains of the three pairs of semicircular canals among the control, unilateral otosclerosis, and

Table 1
Clinical and demographic characteristics of the sample (n=24)

	Otosclerosis (n=12)	Control (n=12)
Female (n, %)	11 (91.6)	11 (91.6)
Age (mean ± SD)	44.4 ± 7.6	44.0 ± 7.9
Bilateral disease (n, %)	6 (50)	-
Cochlear otosclerosis (n, %)	0 (0)	-

The results are expressed as (n, %) for the categorical variables and (mean ± SD) for the continuous variables.

Table 2
Comparative analysis of vHIT results

	Otosclerosis (n=12)	Control (n=12)	*p*
VOR Gain Horizontal SCC (R)	0.97 ± 0.07	1.04 ± 0.22	0.89
VOR Gain Horizontal SCC (L)	0.90 ± 0.07	0.97 ± 0.20	0.76
VOR Gain Anterior SCC (R)	1.05 ± 0.33	0.91 ± 0.09	0.32
VOR Gain Anterior SCC (L)	0.85 ± 0.13	0.91 ± 0.08	0.30
VOR Gain Posterior SCC (R)	0.80 ± 0.09	0.86 ± 0.08	0.06
VOR Gain Posterior SCC (L)	0.86 ± 0.22	0.83 ± 0.10	0.21
Presence of corrective saccades	4 (33.33)	2 (16.67)	0.32

vHIT – Video Head Impulse Test; VOR – vestibulo-ocular reflex; SCC – semicircular canal; R – right; L – left; The gain values are expressed as (mean ± SD), and the presence of corrective saccades is expressed as (n, %).

Table 3
Comparative analysis of DHI results

	Otosclerosis (n=12)	Control (n=12)	*p*
Mean Total Scores	24.08	6.25	0.007*
Physical Domain	6.25	2.33	0.02*
Functional Domain	8.42	2.67	0.006*
Emotional Domain	9.42	1.25	0.003*

DHI – Dizziness Handicap Inventory; The presented values correspond to simple arithmetic means; * significantly different values between the groups, assuming a cutoff of $p<0.05$ to define statistical significance.

bilateral otosclerosis subgroups. There was no significant difference in the mean gains among the three subgroups for any of the canals (Kruskal-Wallis $p>0.05$) or between each pair of two subgroups (Man-Whitney $p>0.05$).
The comparison of the mean gains between the right and left semicircular canals of the sample showed significantly higher values on the right side (Wilcoxon $p<0.001$); no analysis was performed at the level of the affected ears.

Comparative analysis of the DHI results
Regarding the DHI, there was a statistically significant difference between the mean total score of the otosclerosis group and that of the control group (24.1 vs. 6.3; Mann-Whitney $p<0.01$). A similar finding was obtained for the three components of the DHI (physical, functional, and emotional). The results of the comparative analysis of the DHI scores are shown in Table 3.

Discussion

Otosclerosis is one of the most common causes of progressive hearing loss in the adult population.[2] Despite its apparent relationship with vestibular symptoms, the objective evidence for vestibular involvement is, per se, scarce, and this has recently been the focus of clinical investigation.[4–6]
The relationship of otosclerosis with vertigo and balance symptoms has been described in the literature.[10–12] One proposed etiology of these symptoms is the otosclerotic involvement of the vestibular organ itself, which is supported by some histopathological evidence.[11,13] Another theory is that this involvement occurs at the level of the vestibular Scarpa's ganglion.[14] However, the reported symptoms may correspond to a concomitant vestibular disease.[15] The literature has reported isolated cases of benign paroxysmal positional vertigo (VPPB) that may be related to the location and extension of otosclerosis,[16] and that appear to be more common in the early postoperative period of surgical trauma.[17]
In the context of more recent studies on this topic, Catalano et al.[4] prospectively analyzed a sample of patients with otosclerosis and compared them to a control group by performing pre- and postoperative evaluations. Although the authors did not report significant differences in the parameters of the vHIT between the two groups, they suggested the role of cVEMPS in the assessment of postoperative vertigo and indicated a possible saccular dysfunction of traumatic etiology due to its proximity to the oval window.
Moreover, Satar et al.[5] presented a similar hypothesis after comparing patients with otosclerosis (previously operated and non-operated groups) with a control group. They found a significant reduction in the gains of the horizontal semicircular canals and a higher prevalence of corrective saccades in the otosclerosis groups.
More recently, Rajati et al.[6] studied this topic more comprehensively using a larger sample. They observed vestibular dysfunction in the otosclerosis group caused by the involvement of the otolithic system, as shown by abnormal oVEMPs and cVEMPs results, as well as the involvement of the semicircular canals, as

demonstrated by the caloric test and vHIT results (in which the gains were significantly lower in the otosclerosis group than in the control group). To the best of our knowledge, no study thus far has performed a subjective assessment of the quality of life related to vestibular symptoms in patients with otosclerosis by comparing them to healthy individuals. In this study, no significant difference was found in the results of the vHIT between the otosclerosis group (unilateral and bilateral disease) and control group—in the gains calculated for the VOR of the semicircular canals and the presence of corrective saccades (Table 2).

Moreover, there was a statistically significant difference in the total score obtained in the DHI questionnaire and the partial scores obtained in its three domains (physical, functional, and emotional) between the two study groups (Table 3).

Thus, the data suggest a relationship between otosclerosis and a higher prevalence of vestibular symptoms, which does not appear to be explained by the results of the vHIT.

These findings are in line with the results of Catalano et al.[4], who reported that there was no effect of otosclerosis on the function of the semicircular canals, as measured by the vHIT. However, these findings diverge from those of two more recent studies on the topic[5,6], which reported the involvement of this system.

The main limitation of this study was the small sample size, which led to a reduction in the capacity to measure effects. Further, the analysis was performed on individuals rather than the affected ears, and this may have affected the statistical analysis—the inclusion of healthy ears in the unilateral otosclerosis group. The determining factor for this limitation was the fact that the device used in the study (ICS Impulse System, GN Otometrics) is a system of monocular acquisition that tends to overestimate the gains on the side where the camera is positioned (right side). The use of a system with binocular acquisition would circumvent this limitation.

A limitation regarding the vHIT was the fact that the exams were performed by more than one operator, with potential technical bias. Another important limitation, which is common to all studies in which answers to questionnaires are evaluated, was the high subjectivity of their interpretation and potential exceptional uncontrollable factors that may influence the responses at the time of completing the questionnaire.

In the future, this issue should be addressed by using larger samples and conducting comprehensive studies on all components of vestibular function to obtain an overall assessment of these patients. Comparative pre- and postoperative evaluations should also be performed to understand the impact of stapedotomy on vestibular function and determine the presence of modifying factors.

Conclusions

In this study, subjective functional impairment of balance was observed in patients with otosclerosis (as was shown in the DHI questionnaire) who did not appear to be associated with a dysfunction of the semicircular canals, as shown by the vHIT. Further studies should be conducted with larger samples to evaluate this issue and assess the remaining components of vestibular function in patients with otosclerosis—through VEMPs and caloric tests.

Conflicts of Interest

The authors declare that there is no conflict of interests regarding the publication of this paper.

Data Confidentiality

The authors declare having followed the protocols in use at their working center regarding patients' data publication.

Protection of humans and animals

The authors declare that the procedures were followed according to the regulations established by the Clinical Research and Ethics Committee and to the 2013 Helsinki Declaration of the World Medical Association.

Funding Sources

This work did not receive any contribution, funding or scholarship.

Availability of scientific data

There are no datasets available, publicly related to this work.

Bibliographic references

1. House JW, Cunningham CD. Otosclerosis. In: Flint P, Haughey B, Lund V, Robbins K, Thomas JR, Lesperance M. et al Cummings Otolaryngology - Head and Neck Surgery. 7th ed. Philadelphia: Elsevier; 2021. p. 2197-2206.
2. Chole RA, McKenna M. Pathophysiology of otosclerosis. Otol Neurotol. 2001 Mar;22(2):249-57. doi: 10.1097/00129492-200103000-00023.
3. Hannley MT. Audiologic characteristics of the patient with otosclerosis. Otolaryngol Clin North Am. 1993 Jun;26(3):373-87. doi:10.1016/S0030-6665(20)30816-1
4. Catalano N, Cammaroto G, Galletti B, Freni F, Nicita RA, Azielli C. et al. The role of cVEMPs and vHIT in the evaluation of otosclerosis and its eventual vestibular impairment: preliminary findings. B-ENT. 2017;13(1Suppl 27):31-36.
5. Satar B, Karaçaylı C, Çoban VK, Ozdemir S. Do otosclerosis and stapedotomy affect semicircular canal functions? Preliminary results of video head impulse test. Acta Otolaryngol. 2021 Apr;141(4):348-353. doi: 10.1080/00016489.2021.1873416.
6. Rajati M, Jafarzadeh S, Javadzadeh R, Disfani MF, Yousefi R. Comprehensive vestibular evaluation in patients with otosclerosis: a case control study. Indian J Otolaryngol Head Neck Surg. 2022 Dec;74(4):582-587. doi: 10.1007/s12070-022-03147-5.
7. Halmagyi GM, Chen L, MacDougall HG, Weber KP, McGarvie LA, Curthoys IS. The video head impulse test. Front Neurol. 2017 Jun 9;8:258. doi: 10.3389/fneur.2017.00258.
8. Garcia FV, Luzio CS, Benzinho TA, Veiga GB. Validação e adaptação do dizziness handicap inventory para a língua e população portuguesa de Portugal. Acta ORL. 2008; 26(2):128-132.
9. Cureoglu S, Baylan MY, Paparella MM. Cochlear Otosclerosis. Curr Opin Otolaryngol Head Neck Surg. 2010 Oct;18(5):357-62. doi: 10.1097/MOO.0b013e32833d11d9.
10. Morales-Garcia C. Cochleo-vestibular involvement in otosclerosis. Acta Otolaryngol. 1972 Jun;73(6):484-92. doi: 10.3109/00016487209138969.
11. Ghorayeb BY, Linthicum FH Jr. Otosclerotic inner ear syndrome. Ann Otol Rhinol Laryngol. 1978 Jan-Feb;87(1 Pt 1):85-90. doi: 10.1177/000348947808700115.
12. Cody DT, Baker HL Jr. Otosclerosis: vestibular symptoms and sensorineural hearing loss. Ann Otol Rhinol Laryngol. 1978 Nov-Dec;87(6 Pt 1):778-96. doi: 10.1177/000348947808700605.
13. Sando I, Hemenway WG, Miller DR, Black FO. Vestibular pathology in otosclerosis temporal bone nistopathological report. Laryngoscope. 1974 Apr;84(4):593-605. doi: 10.1288/00005537-197404000-00012.
14. Saim L, Nadol JB Jr. Vestibular symptoms in otosclerosis - correlation of otosclerotic involvement of vestibular apparatus and Scarpa's ganglion cell count. Am J Otol. 1996 Mar;17(2):263-70.
15. Morozova SV, Dobrotin VE, Kulakova LA, Kaspranskaia GR, Ovchinnikov IuM. Vestibular disorders in patients with otosclerosis: prevalence, diagnostic and therapeutic options. Vestn Otorinolaringol. 2009;(2):20-2.
16. Crossland G, De R, Axon P. Far advanced otosclerosis and intractable benign paroxysmal positional vertigo treated with combined cochlear implantation and posterior semicircular canal occlusion. J Laryngol Otol. 2004 Apr;118(4):302-4. doi: 10.1258/002221504323012076.
17. Atacan E, Sennaroglu L, Genc A, Kaya S. Benign Paroxysmal Positional Vertigo After Stapedectomy. Laryngoscope. 2001 Jul;111(7):1257-9. doi: 10.1097/00005537-200107000-00021.