

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Floating labyrinth: A unique finding on CT scan ☆☆☆

Filipe Correia, M.D.^{a,b,*}, Rita Sousa, M.D.^{a,b}, Jorge Domingues, M.D.^{a,b}^a Serviço de Otorrinolaringologia, Hospital de Egas Moniz, Centro Hospitalar de Lisboa Ocidental, Rua da Junqueira 126 1349-019, Lisboa, Portugal^b Nova Medical School, Universidade Nova, Campo Mártires da Pátria 130 1169-056, Lisboa, Portugal

ARTICLE INFO

Article history:

Received 21 June 2020

Revised 8 August 2020

Accepted 9 August 2020

Keywords:

Floating labyrinth

Chronic suppurative otitis media

Labyrinthitis

AIDS

CT scan

Bony sequestrum

ABSTRACT

We report a case of a 67-year-old female patient that presented to the emergency department with complete right facial paralysis, progressive hearing loss and chronic otorrhea. A unique finding on CT scan is reported: a “floating labyrinth” – cochlea, vestibule and semicircular canals extensively surrounded by soft tissue density material but with intact thinned otic capsule walls. A transotic approach was performed for removal of noncholesteatomatous inflammatory tissue; intravenous antibiotics and corticosteroids led to partial recovery of facial nerve function. A chronic suppurative otitis media with necrotic osteomyelitis and bony sequestrum in a severe context of AIDS is the likely cause. An immunosuppressive disease should be suspected in atypical presentations of chronic suppurative otitis media without cholesteatoma.

© 2020 The Authors. Published by Elsevier Inc. on behalf of University of Washington.

This is an open access article under the CC BY-NC-ND license.

<http://creativecommons.org/licenses/by-nc-nd/4.0/>

Introduction

Although chronic otitis media is a common finding in our daily practice, labyrinth erosion has become a rare finding in the developed world. It is usually detected on ear CT scan as a lateral semicircular canal fistula, but a complete erosion of posterior labyrinth has occasionally been reported because of cholesteatoma. We describe the case of a patient with a unique finding on CT scan, caused by a massive inflammatory erosion of the inner ear.

Case report

A 67-year-old woman presented to the otorhinolaryngology emergency department with a 6-month history of progressive right facial nerve paralysis, progressive hearing loss and chronic otorrhea of the right ear. A grade VI facial paralysis (House-Brackman classification) was evident and an obliterative inflammatory-like mass was detected on the right external auditory canal. Temporal bone-CT-scan (Fig. 1) showed an extensive infiltrate occupying middle ear with

☆ Local where the work was carried out: Hospital de Egas Moniz, Rua da Junqueira 126, 1349-019, Lisboa, Portugal.

☆☆ Competing Interests: There are no financial conflicts of interest to disclose.

* Corresponding author.

E-mail address: fcorreia@chlo.min-saude.pt (F. Correia).<https://doi.org/10.1016/j.radcr.2020.08.021>1930-0433/© 2020 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license. (<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

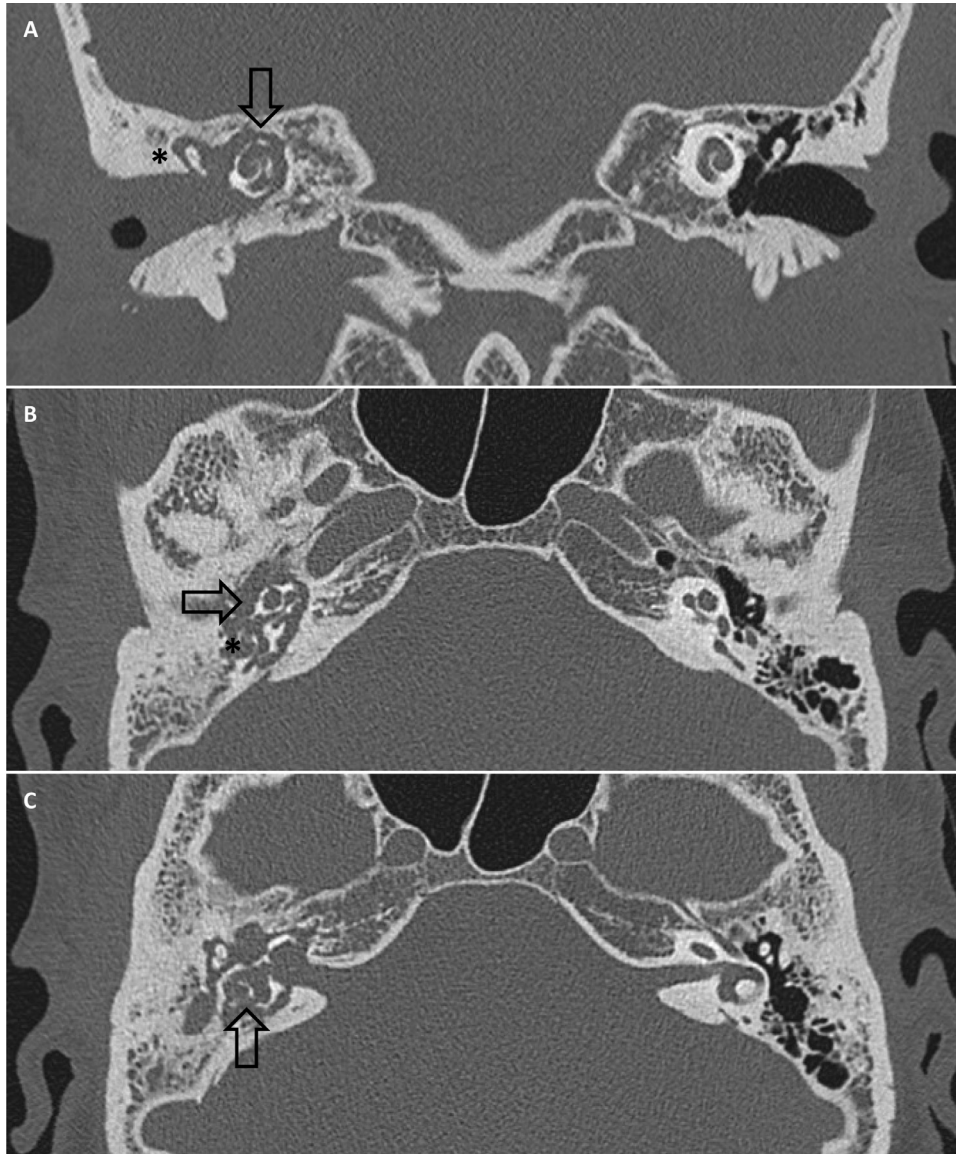


Fig. 1 – (A-C): Floating labyrinth on CT scan: (A) (coronal CT image bone window): extensive soft tissue density infiltrate involving right external auditory canal, middle and inner ear; scutum intact (*); suspected erosion of malleus handle, long process of incus and stapes; otic capsule erosion, but its contour, thinned, and its turns stand clearly delineated (arrow), giving this aspect of “floating labyrinth”. (B) (axial CT image bone window): complete mastoid air cells obliteration; apex and middle turn of cochlea (arrow) and posterior semicircular canal ampulla are identified; facial nerve (*) turns identifiable in its mastoid segment. (C) (axial CT image bone window): vestibule and lateral semicircular canal identified (arrow); its dome is unexpectedly preserved.

ossicular chain erosion and complete mastoid air cells obliteration, extending into the external auditory canal and eroding inner ear. CT scan findings of the inner ear were impressive: many components of the labyrinth were clearly identifiable but surrounded by soft tissue density material; the otic capsule retained its contour but its thickness was severely reduced; the facial nerve pathway is lost in the middle ear. Patient was treated with systemic corticosteroids, endovenous antibiotics, and urgent surgery. All inflammatory tissue was removed until the internal auditory canal was reached.

Granulation tissue and necrotic cochlear bone fragments are clearly seen in intraoperative pictures (Fig. 2A). Histopathologic analysis showed inflammatory granulation tissue and eroded compact bone with no evidence of cholesteatoma (Fig. 2B). A grade IV facial paralysis (House-Brackman classification) and a non-discharging ear was achieved 6 months after surgery. Secondary study revealed a chronic HIV and HBV infection with severe immunosuppression. Syphilis and tuberculosis have been ruled out by serologic and culture tests, respectively.

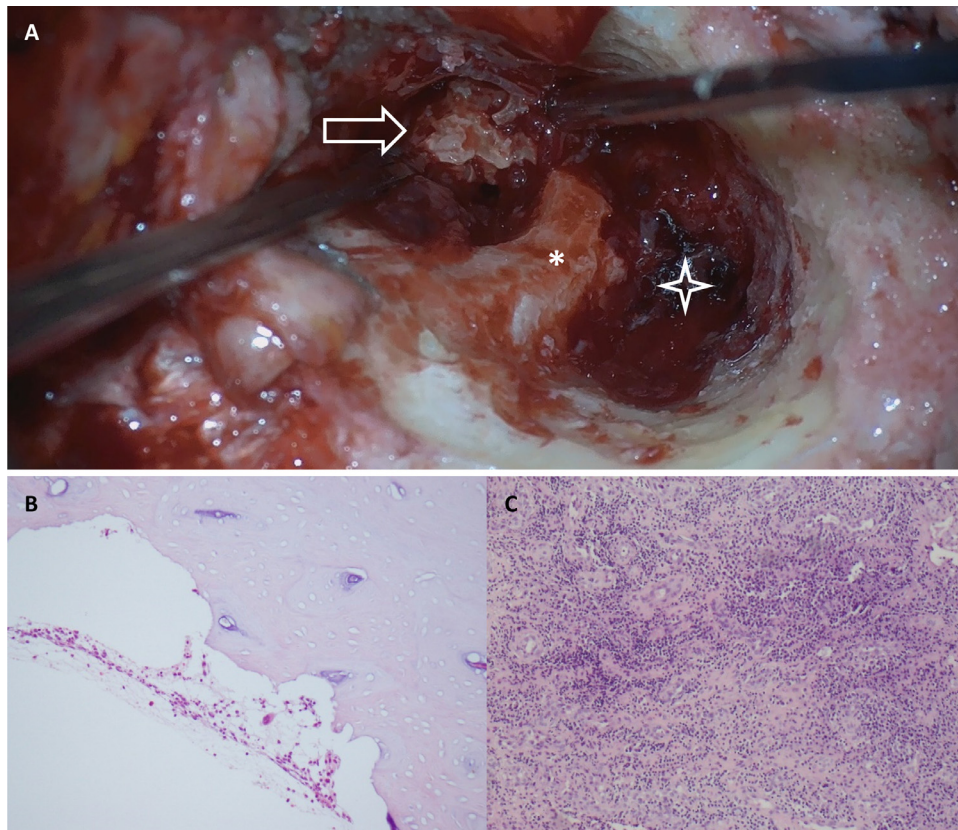


Fig. 2 – (A-B) Intraoperative and histopathological findings; (A) intraoperative removal of cochlea; a canal-wall-down mastoidectomy was performed (★), with a progressive removal of inflammatory tissue and necrotic bone debris; cochlea floating in inflammatory tissue was removed piece by piece (arrow); the facial nerve was identified and preserved (* facial ridge). (B) (HE 100x): eroded compact bone (otic capsule) surrounded by inflammatory cells. (C) (HE 100x): exuberant granulation tissue: lymphocytes and some neutrophils surround large areas of new blood vessels in a loose extracellular matrix.

Discussion

Tympanogenic inner ear erosion usually affects the lateral semicircular canal and is invariably related to cholesteatomatous chronic otitis media [1-3]. Labyrinthine fistula develops as consequence of slowly progressive erosion of the bony labyrinth in 2.7%-12.5% of cholesteatomatous ears [4-7] but can also occur associated chronic otitis media with granulation [3]. Vertigo is usually the presenting symptom but patients may remain asymptomatic for years [6,7]. A suppurative labyrinthitis may also develop and an insidious clinical course of hearing loss and disequilibrium in a chronically discharging ear is possible [6,8].

Imaging and pathologic changes of suppurative labyrinthitis tend to occur in inner ear spaces, more than in the otic capsule itself [2]. However, anecdotal reports of extensive bony destruction in the setting of suppurative labyrinthitis have been described, with inner ear diffusely destroyed by inflammatory granulation tissue and bony sequestrum

by necrotic osteomyelitis [9]. Bony sequestrum represents devitalized bone that has become separated from the surrounding bone because of infectious necrosis (usually in a setting of osteomyelitis, tuberculosis or tumor); on CT scan a piece of calcified tissue is noticeable within a lucent lesion [10,11].

In recent reports, HIV and other immunosuppressive settings have been reported as predisposing factors for osteomyelitis of the temporal bone associated to necrotizing otitis externa [12,13].

This case demonstrates a unique form of suppurative labyrinthitis, where the inner ear was progressively involved and destroyed by an extensive inflammatory suppurative reaction. Otic capsule walls were progressively thinned but preserved its morphology on CT scan, giving this aspect of “floating labyrinth”. A chronic necrotic osteomyelitis with bony sequestrum in a severe immunosuppressive context is the likely cause; this etiology should be suspected in atypical presentations of chronic suppurative otitis media without cholesteatoma.

REFERENCES

- [1] Lemmerling MM, De FB, Verbist BM, VandeVyver V. Imaging of inflammatory and infectious diseases in the temporal bone. *Neuroimag Clin N Am* 2009;19(3):321–37. doi:10.1016/j.nic.2009.06.006.
- [2] Paparella MM, Sugiura S. The pathology of suppurative labyrinthitis. *Ann Otol Rhinol Laryngol* 1967;76:554–86.
- [3] Smith JA, Danner CJ. Complications of chronic otitis media and cholesteatoma. *Otolaryngol Clin North Am* 2006;39:1237–55. doi:10.1016/j.otc.2006.09.001.
- [4] Misale P, Lepcha A, Chandrasekharan R, Manusrut M. Labyrinthine fistulae in squamosal type of chronic otitis media : therapeutic outcome. *Iran J Otorhinolaryngol* 2019;3(10):167–72.
- [5] Sanna M, Zini C, Bacciu S, Scandellari R, Delogu P, Jemmi G. Management of the labyrinthine fistula in cholesteatoma surgery. *ORL* 1984;172:165–72.
- [6] Schmidt LP, Canali I, Teixeira A, Noschang M, Selaimen F, Selaimen S. Cholesteatoma labyrinthine fistula : prevalence and impact. *Braz J Otorhinolaryngol* 2018(xx):1–6. doi:10.1016/j.bjorl.2018.01.005.
- [7] Grewal DS, Hathiram BT, Dwivedi A, Kumar L, Sheth K, Srivastava S. Labyrinthine stula : a complication of chronic suppurative otitis media. *J Laryngol Otol* 2003;117:353–7.
- [8] Maranhão AS, de A, Godofredo VR, Penido N, de O. Suppurative labyrinthitis associated with otitis media: 26 years' experience. *Braz J Otorhinolaryngol* 2016;82(1):82–7. doi:10.1016/j.bjorl.2014.12.012.
- [9] Lao Z, Sha Y, Dai CF, Huang WH, Cheng YS. End-stage labyrinthitis with extensive erosion of the bony labyrinth. *Otol Neurotol* 2012;33(7):53–4. doi:10.1097/MAO.0b013e318248ee99.
- [10] Jennin F, Bousson V, Parlier C, Jomaah N, Khanine V, Laredo J-D. Bony sequestrum : a radiologic review. *Skelet Radiol* 2011;40:963–75. doi:10.1007/s00256-010-0975-4.
- [11] Helms CA, Jeffrey RB, Wing VW. Skeletal radiology computed tomography and plain film appearance of a bony sequestration : significance and differential diagnosis. *Skelet Radiol* 1987;16:117–20.
- [12] Prasad SC, Prasad KC, Kumar A, Thada ND, Rao P, Chalasani S. Osteomyelitis of the temporal bone: terminology, diagnosis, and management. *J Neurol Surgery Part B Skull Base* 2014;75(5):324–31. doi:10.1055/s-0034-1372468.
- [13] Ayueyda J, Thomas DM, Main J, Patel KS. Malignant otitis externa in HIV and AIDS. *J Laryngol Otol* 1996;110:770–5.