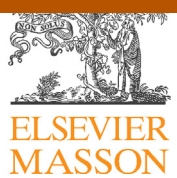
European Annals of Otorhinolaryngology, Head and Neck diseases (2013) 130, 341—343



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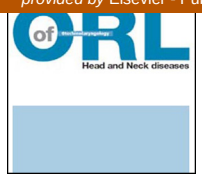

CASE REPORT

Spontaneous osteo-dural fistulae of petrous bone posterior wall

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KEYWORDS
Arachnoid granulation;
Cerebrospinal fistula;
Temporal bone

Summary
Objective: To raise awareness of the possibility of spontaneous temporal bone cerebrospinal fistula in case of clear retrotympanic effusion.
Case report: A 63-year-old man with no particular history presented with unilateral spontaneous right retrotympanic clear effusion. CT found defects in the posterior part of the right temporal bone, in contact with arachnoid granulations, with no other visible abnormalities.
Discussion/Conclusion: Unilateral clear retrotympanic effusion in an adult subject should, apart from serous otitis media, suggest possible cerebrospinal fistula. In the absence of otologic or traumatic history, arachnoid granulation is one possible etiology, inducing spontaneous cerebrospinal fluid leakage when facing the temporal bone. Diagnosis is suggested by bone defects in the tegmen tympani or posterior wall of the temporal bone on CT, with the adjacent mastoid cavities filled with fluid. Pneumococcal vaccination and early surgical repair of the fistula should be performed to avoid neuromeningeal infection.

Introduction

In case of unilateral retrotympanic effusion in adults without otologic history, the ENT physician should investigate etiology if ipsilateral nasopharyngeal tumor has been ruled out [1]. One possible cause to be considered in absence of old or recent trauma is spontaneous cerebrospinal fistula of the temporal bone, due either to associated meningocele or meningo-encephalocele or to ectopic arachnoid granulation [2].

Arachnoid granulations drain and absorb cerebrospinal fluid (CSF) in the venous sinuses [1—3], but are sometimes ectopic, without communication with the venous sinuses and may, with time, induce erosion by continuous pulsatile CSF pressure on the cortex of the adjacent bone, creating a fistula with spontaneous CSF leakage into the facing anatomic cavities [1—4].

We report a case of persistent spontaneous clear retrotympanic effusion.

Case report

A 63-year-old man presented with right hearing loss and a blockage of the right ear that he had been feeling for 1 month. There was no history of trauma or of temporal bone or middle ear infection. He also reported some recent episodes of salty-tasting posterior rhinorrhea. Otoscopy found spontaneous right retrotympanic clear effusion

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1879-7296/$ - see front matter © 2013 Published by Elsevier Masson SAS.
http://dx.doi.org/10.1016/j.anorl.2012.09.013

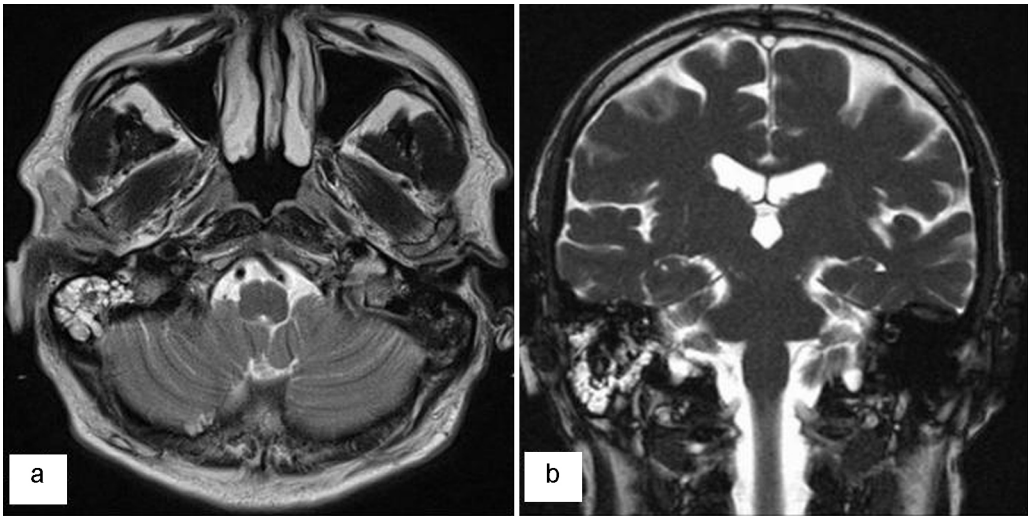

Figure 1 a: posterior fossa MRI, T2-weighted axial slice: fluid filling of right mastoid cells; b: coronal slice: absence of associated meningocele or meningo-encephalocele.

with a normal tympanum. Urine strip test after Dandy maneuver indicated rhinoliquorrhea.

There were no signs of intracranial hypertension. Tonal audiometry found right transmission hearing loss (−25 dB at 1000 Hz).

Millimetric axial and coronal temporal bone CT slices found a defect on the posterior side of the ipsilateral temporal bone, without meningocele or meningo-encephalocele or other associated morphologic abnormalities (Fig. 1).

The left side showed no particularities.

Sinus CT was normal. Cerebellopontine angle MRI found fluid filling the right mastoid cells, with no associated lesions (Figs. 2a, b and 3) or signs of intracranial hypertension. Comparison of clinical and radiological findings indicated a spontaneous osteodural fistula with bone lysis of the posterior wall of the right temporal bone, secondary to ectopic arachnoid granulation.

Mastoidectomy was successfully performed in June 2011, on a retro-auricular approach, closing the fistula with autologous bone powder and biologic glue. Pneumococcal vaccination was performed to prevent bacterial meningitis.

Discussion

Etiologically, osteodural fistula with clear retrotympanic effusion is related to continuity of the temporal pyramid at the middle cranial fossa, usually involving the tegmen tympani. Mechanisms may be inflammatory (chronic otitis media with or without cholesteatoma), traumatic, iatrogenic (otologic surgery), tumoral or congenital.

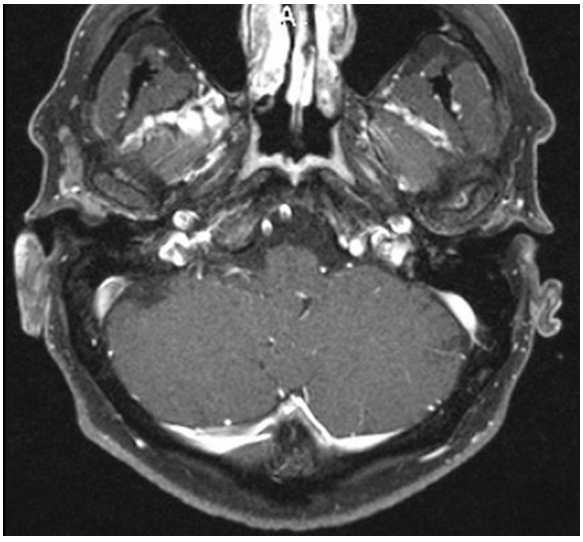

Figure 2 Posterior fossa MRI, T1-weighted gadolinium-enhanced slice: absence of lesion of the posterior side of the temporal bone (endolymphatic sac tumor or meningioma); absence of uptake in facing meninx.

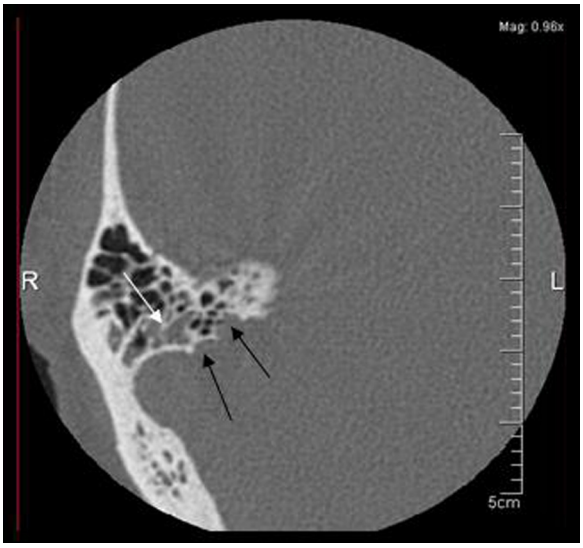

Figure 3 Right temporal bone CT, axial slice, inverted contrast: pre-sinus bone defect of the posterior side of the temporal bone (black arrows), with limited superficial focal aspect indicative of erosion, related to arachnoid granulation. Partial filling of facing mastoid cells (white arrows).

Posterior involvement of the temporal pyramid is more rare, and usually congenital [2,4]. There may also be temporal bone erosion by ectopic arachnoid granulation, in which case involvement may be uni- or bilateral and isolated or multiple. Gender and ethnicity are not factors [2,4]; age is the main predictor of onset, which is at a mean age of 55 years [4] with prevalence rising to 28% in over 50-year-old [2].

Spontaneous rhinoliquorrhea shows similar demographic patterns, and there is a strong clinical and radiological correlation with idiopathic intracranial hypertension [5].

A study of temporal bone sections found arachnoid granulation eroding the temporal pyramid, without associated abnormalities, in 13% of cases, half of which concerned the posterior side of the bone [2], whereas incidence of spontaneous effusion related to CSF fistula was much lower. This suggests that arachnoid granulation alone is not enough to induce clinically visible otoliquorrhea and that physiopathologic factors are involved: excess weight, reduced bone density (infection, malnutrition), congenital bone thinning, and intracranial hypertension [2].

Clinically, there is clear retrotympanic effusion with a normal tympanum, and transmission hearing loss on tonal audiometry. Persistent clear otorrhea observed on paracentesis and/or implantation of transtympanic aerators for suspected serous otitis media [1] may be revelatory in absence of any otologic history [6]. Diagnosis of otoliquorrhea is confirmed, in case of doubt, by beta-2-transferrin found on immuno-electrophoresis [4,6]. Labstix screening for glucose yields as many as 20 to 30% false positives. Dandy maneuver should be systematic, to explore for clear rhinorrhea [3], corresponding to rhinoliquorrhea via the auditory tube.

Investigation of intracranial hypertension is mandatory, especially in case of the above physiopathological criteria. CSF opening pressure should be measured by lumbar puncture, being a risk factor for postoperative recurrence [5,7].

Diagnosis is suggested on millimetric axial and coronal temporal bone CT slices [3]. The clinician is alerted by fluid filling the mastoid cavities. Defects should be looked for over the entire temporal bone, and especially at the tegmen tympani, the posterior side and at the anterior sinus, with a limited superficial focal aspect indicative of erosion [6]. Certain frequently associated lesions such as meningocele and meningoencephalocele should be looked for. Other causes of temporal bone osteodural fistula should be ruled out: superior semicircular canal dehiscence, cerebellopontine angle meningioma or endolymphatic sac tumor; the latter two are ruled out on MRI (T1-weighted gadolinium-enhanced sequence), which also confirms presence of CSF in the mastoid cells (T2-weighted sequence) [4,8] and can rule out signs of intracranial hypertension (empty sella turcica, ventricular dilation, signs of transependymal resorption: FLAIR sequence).

Conclusion

Unilateral spontaneous clear retrotympanic effusion in absence of otologic history or of cranial trauma should suggest possible osteodural fistula. Early treatment by surgical filling and preventive pneumococcal vaccination avoids potentially serious meningeal infection [1,6].

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

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