Non-Traumatic Epitympanic Cerebrospinal Fluid Leakage Presenting as Recurrent Bacterial Meningitis

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Recurrent bacterial meningitis is a medical problem more frequently encountered in children than adults [1]. When it occurs in adults it is mainly due to cerebrospinal fluid leak associated with closed head trauma or neurosurgical complications.

Recurrent bacterial meningitis in a previously healthy young adult without any history of CSF leak or closed head trauma is challenging. We describe here the clinical course and the radiological and laboratory parameters of a healthy young adult with a normal tympanic otoscopy who suffered from otogenic recurrent bacterial meningitis.

PATIENT DESCRIPTION

A 23 year old man was admitted after a day of fever and headache. On admission his conditioned worsened. The patient was comatose and his neurologic examination demonstrated severe nuchal rigidity. Brain computed tomography scan did not reveal any pathologic findings. Lumbar puncture suggested bacterial meningitis. He was treated with intravenous ceftriaxone and his condition rapidly improved. He completed a 10 day course of ceftriaxone. Blood and CSF cultures were negative. His medical history was unremarkable; there was no family history of recurrent invasive bacterial infections.

Eight weeks after discharge the patient was readmitted again with acute meningitis. Brain CT scan showed diffuse edema. After blood cultures were obtained he was treated with intravenous ceftriaxone. A lumbar puncture showed marked neutrophilic leukocytosis, increased protein levels and decreased glucose levels. The patient's condition improved dramatically during 24 hours of treatment. Blood and CSF cultures were negative. A repeat lumbar puncture after 2 weeks of antibiotic treatment showed 16 white blood cells/mm^3, increased protein level (127 g/dl) and decreased glucose level (35 mg/dl) (blood glucose level was 90 mg/dl)

Given the extremely unusual course of two consecutive episodes of presumably bacterial meningitis (in view of the rapid response to antibacterial therapy), an extensive workup was carried out. Quantitative immunoglobulin and complement levels and activity (including terminal complement component) were within normal limits. Human immunodeficiency virus test was negative. Brain magnetic resonance imaging and high resolution CT showed opacification of the left mastoid air cells and middle ear of the same side [Figure A] with a suspected bone erosion of the tegmen tympani [Figure B], normal width of the internal acoustic canal, and absence of encephalocele. Polymerase chain reaction was performed on a CSF sample for a gene fragment of the 16S rRNA. The PCR product was sequenced and the nucleotide sequence was determined and compared with the National Center for Biotechnology Information database using the Basic Local Alignment Search Tool (BLAST). It showed a 99% DNA sequence homology with *Streptococcus mitis*.

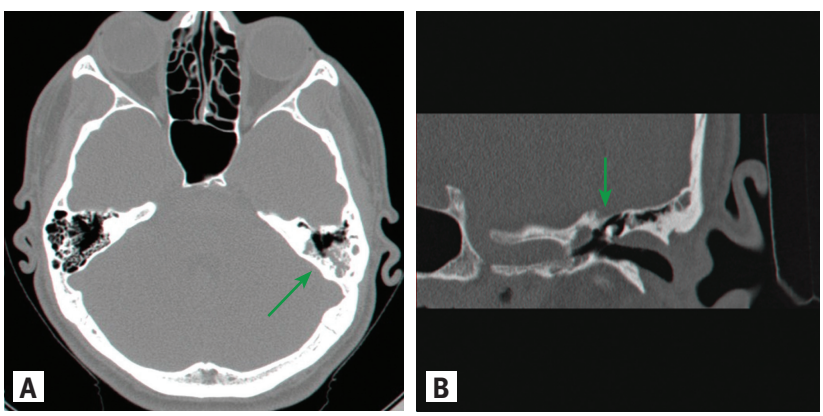
The patient was referred to the otolaryngology surgeons. Microscopic oto-

CSF = cerebrospinal fluid

PCR = polymerase chain reaction

[A] Axial CT scan showing opacification of the left mastoid air cells

[B] Coronal CT scan showing tegmen defect (green arrow)



scopy of the ear showed mild edema of the distal canal at the posterior wall. The tympanum was intact and clear. He was treated with intravenous ceftriaxone until a diagnostic exploration of the mastoid could be conducted. An explorative tympanomastoidectomy was performed through a retro-auricular incision; a large fascial section was collected from the temporalis fascia. No pathology was found on the lateral semicircular canal (mastoid cavity) or in the area of the round and oval window (tympanic cavity), but a watery leak was spotted in the facial recess emerging from the tympanic attic (the superior wall or roof of the tympanic cavity). A wide atticotomy was performed and a defect was found in the tegmen with CSF leakage seen clearly emerging from a dural defect. The defect was sealed using temporalis fascia covered by muscle and then bone wax. The patient is still in follow-up, without any signs of infection.

COMMENT

This unusual case of recurrent bacterial meningitis was caused by a bony defect in the tegmen tympani (epitympanic dehiscence), a part of the temporal bone. Spontaneous CSF leak into the middle ear is rare and can easily be missed. The leak can occur without any previous history of head trauma or temporal bone fractures. Furthermore, autopsy studies suggest that single defects in the tegmen of the temporal bone occur in 15% to 34% and multiple defects in less than 1% [2,3]. We suspect that the recent increase in reported cases represents an increased recognition of the disease process. The onset of meningitis only at the age of 23 is probably related to the timing of the dural rupture. The presenting symptom of these defects with intact ear drum is usually rhinorrhea, and when left untreated could cause meningitis.

Otorrhea due to epitympanic dehiscence and meningitis have been described previously, but to the best of our knowledge this is the first reported case of a tegmen tympani defect presenting as recurrent bacterial meningitis with a normal otoscopy and without a history of otorrhea, trauma, surgery or chronic otitis media. The causative organism was *Streptococcus mitis*, a commensal of the oral flora that caused contamination of the CSF. The case presented reaffirms the notion that recurrent bacterial meningitis should always prompt a search for an underlying cause [4]. Given the lack of suggestive clues from his medical history and owing to a high index of suspicion for a fistula or dehiscence especially when the patient's tympanum was intact and only an opacification of the mastoid was seen, an explorative tympanomastoidectomy seemed mandatory.

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