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Case Report

Unruptured translabyrinthine meningocele without CSF otorrhea

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ABSTRACT

Labyrinthine meningocele can be classified into translabyrinthine and perilabyrinthine type. We describe a case of rare unruptured translabyrinthine meningocele (TLM). It is rare to encounter an unruptured TLM because it is usually diagnosed after rupture as a labyrinthine fistula, cerebral spinal fluid otorrhea, and subsequent meningitis. We provide for the first time an intraoperative photo and video of a case of an unruptured TLM that developed through an inner ear malformation in a single-side deaf child, which was preoperatively misdiagnosed as congenital cholesteatoma in preoperative temporal bone computed tomography. TLM without CSF otorrhea in an unruptured state merit attention because of its importance during the workup of congenital cholesteatoma or cochlear implantation in spite of its rarity of reports.

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1. Introduction

Labyrinthine meningocele is a rare disease formed by an arachnoid membrane that herniates through defects close to the labyrinth, which can be misdiagnosed during the differential diagnosis of the middle ear lesion. We present a case of translabyrinthine meningocele (TLM) that was preoperatively misdiagnosed as congenital cholesteatoma.

2. Case. Translabyrinthine meningocele with a vestibulocochlear malformation presenting as deafness and a middle ear mass

A 7-year-old girl visited our otology clinic to receive surgical treatment for congenital cholesteatoma. She had a history of facial nerve paralysis at 2 years old and a brief seizure attack 2 years ago. The otomicroscopic and otoendoscopic findings suggested a round mass under the intact tympanic membrane in the posterosuperior quadrant, suggesting congenital cholesteatoma. However, she had a complete loss of hearing (deafness) or sensorineural hearing loss (SNHL), which was not a finding of congenital cholesteatoma that could be diagnosed during conductive hearing loss. Although she did not complain of dizziness, she showed no response to caloric stimulation of her left ear.

A high-resolution computed tomographic (CT) examination (1.3 mm-thick sections, Discovery CT750 HD, U.K) through the temporal bone in the axial and coronal planes revealed a dysplastic cochlea without separate apical and second turns and the absence of a normal modiolus; short, abnormal lateral and posterior semicircular ducts; and a broad connection with a large vestibule (Fig. 2). These features correlated with Mondini's dysplasia or incomplete partition type I (IP-I) cystic cochleovestibular malformation acongenital cholesteatomaording to the classification described by Sennaroglu and Saatci [1]. The opening between the cochlea and the deep internal auditory canal (IAC) revealed a possible connection between the cerebrospinal fluid and the labyrinthine fluid.

There was additionally an oval soft tissue mass in the middle ear that was compatible with the otoendoscopic finding of a round mass under the tympanic membrane. The incudostapedial joint was destroyed with an intact malleus and incus, which was consistent with congenital cholesteatoma abutting the incudostapedial joint. There was neither any fluid collection nor facial canal enlargement suggestive of cerebrospinal fluid otorrhea. The congenital malformation of the inner ear and middle ear mass did not seem to be connected in the axial section, whereas there was a suspected connection in the coronal section.

Exploratory tympanotomy was performed under general anesthesia via an endaural incision to confirm the diagnosis, check the CSF otorrhea and remove the middle ear mass if it was congenital cholesteatoma. After a Koener flap and tympanomeatal flap elevation, a thin membrane-walled middle ear cyst was found behind the malleus handle in the area of the oval window. It was not congenital cholesteatoma, and the arachnoid membrane-like

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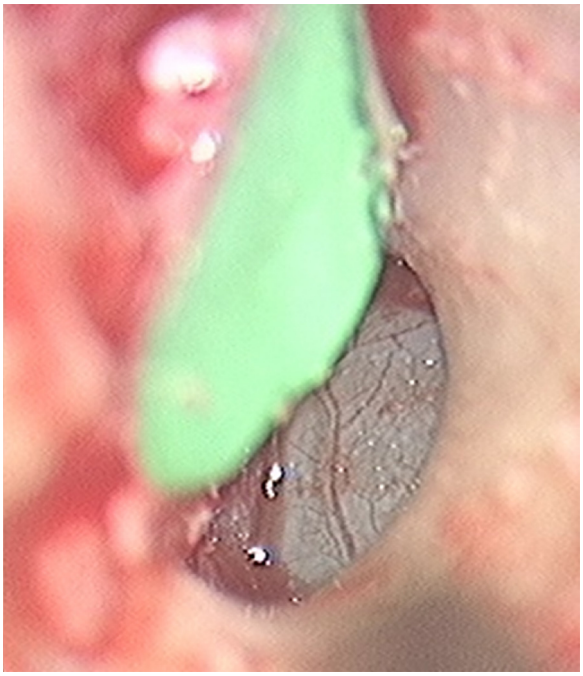

Fig. 1. Translabyrinthine meningocele. F/7 years. Intraoperative photo of after tympanomeatal flap elevation. A pulsatile left middle ear cyst covered with a thin, partially transparent arachnoid membrane and filled with clear fluid was found and intraoperatively diagnosed as translabyrinthine meningocele. The meningocele was protected using a perichondrium graft from the conchal cartilage.

thin wall of the cyst and clear fluid contents of the middle ear mass led to a diagnosis of temporal bone meningocele. The diagnosis of meningocele was made intraoperatively (Fig. 1; the operation video is also provided in the supplemental material.). The oval window and stapes located in the center of mass in the coronal section of the temporal bone CT scan indicated that the meningocele belonged to the translabyrinthine type. Extreme care was taken to avoid rupturing the meningocele, and it was not aspirated because of the risk of immediate profuse CSF otorrhea and later meningitis that would require subtotal petrosectomy (neurotologic surgery and closure of the external auditory canal). It was confirmed that there was no CSF leakage in the middle ear, and the meningocele was not ruptured and showed no surgical trauma. The unruptured meningocele was covered with perichondrium and cartilage composite graft obtained from conchal cartilage to protect it from later trauma and to prevent CSF leakage (Fig. 2).

Postoperatively, the child did not present any vertigo, headache or signs of CSF otorrhea. Postoperative otoendoscopy was submitted in the supplemental video. A one-month postoperative T2-weighted magnetic resonance (MR) (Siemens, Erlangen, Germany, 3.0T) (Fig. 3) revealed a less than 5-mm (slightly decreased size) known meningocele within the left middle ear cavity (asterisk) connected to the cystic semicircular canal, as well as vestibular dysplasia and cochlea dysplasia (white arrowhead). The IAC and the vestibulocochlear malformation did not show a direct connection, but the true IAC of the Y-shaped IAC (white arrow) was observed connecting the inner ear cavity with the peripontine cistern. A pediatric neurosurgeon was consulted to perform transmastoid removal of the meningocele, but regular follow-up was planned instead of impending surgical treatment. Parental education focused on the possibility of meningitis or cerebrospinal fluid otorrhea, which could be potentially life threatening and require subtotal petrosectomy.

3. Discussion

Our case was not present with CSF otorrhea, but the classification of temporal bone meningocele generally follows the classification of CSF otorrhea [2]. Spontaneous CSF otorrhea is a rare but potentially life-threatening condition with two different subtypes: (1) translabyrinthe type in children with inner ear malformation and SNHL, and (2) perilabyrinthine type in adults with meningoencephaloceles [3]. Reported cases include 80 cases of childhood-type translabyrinthine fistula by 2002 [4], 57 cases of

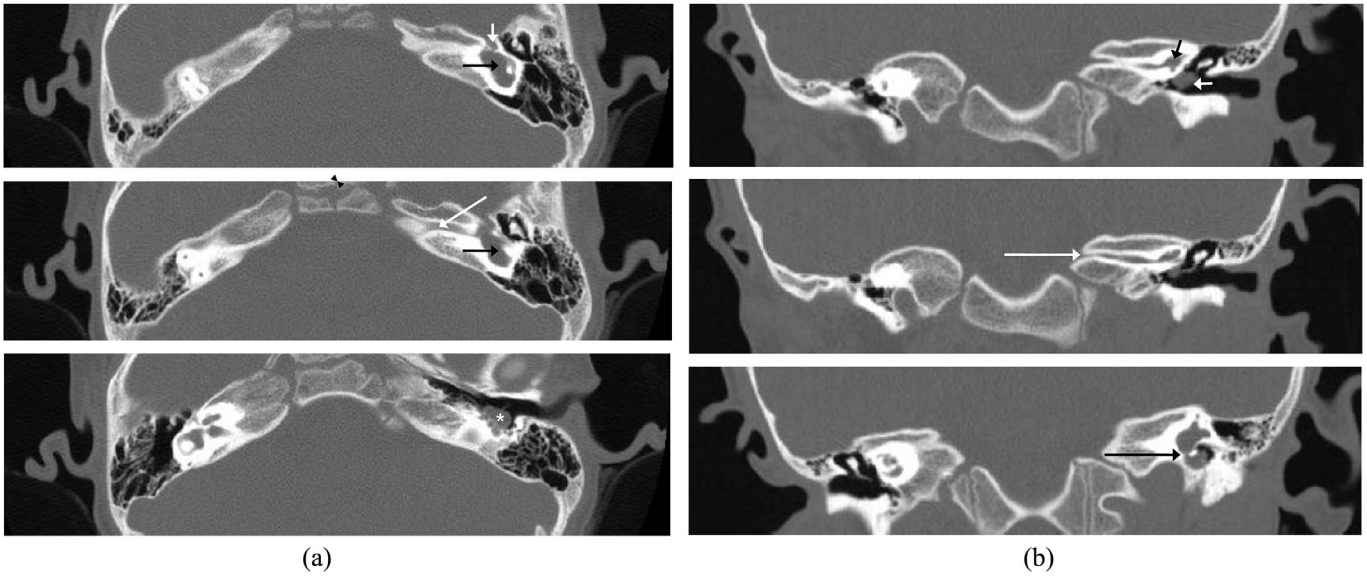

Fig. 2. (a) Axial high-resolution CT images reveal an oval soft tissue mass (*asterisk*) in the middle ear near the oval window that was caudal to the inner ear malformation (*black arrow*), indicating a cystic cochleovestibular malformation incomplete partition type I (IP-I) and a geniculate ganglion opening of the facial nerve (*short white arrow*). The *Y*-shaped internal auditory canal (*long white arrow*) was observed. The incudostapedial joint was destroyed with an intact malleus and incus, which was consistent with congenital cholesteatoma abutting the incudostapedial joint. There was neither any fluid collection nor facial canal enlargement suggestive of cerebrospinal fluid otorrhea. (b) Coronal high-resolution CT images show a 7 mm × 4 mm × 3 mm opacification (*short white arrow*) in the left mesotympanum posterior to the malleus and anterior to the incus abutting the incudostapedial joint and cochlear promontory, of which the first differential diagnosis was congenital cholesteatoma arising from the incudostapedial joint. The internal auditory canal is connected to the vestibule (*black arrow*) and narrowed (*long white arrow*). The posterior semicircular canal dysplasia is connected to a translabyrinthine meningocele (*long black arrow*).

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