

Case Report

Recurrent Meningitis in an 11-year-old Girl with a Petrous Apex Cystic Lesion

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Cases of recurrent meningitis in elderly patients with a spontaneous cerebrospinal fistula have been reported, and in some of these patients, cystic lesions were thought to be the underlying cause. We report a case of recurrent meningitis in an 11-year-old Japanese girl with an arachnoid cyst in the petrous apex. Pulsation of the cystic lesion was thought to cause bone erosion, leading to the formation of a fistula. Magnetic resonance imaging was useful in evaluating the arachnoid cyst and fistula. During 2 years of follow-up, the osteolytic lesion enlarged and the rate of bone erosion was higher than expected.

Key words: magnetic resonance imaging, pulsation of cystic lesion, bone erosion, fistula

Bacterial meningitis usually occurs in patients younger than 2 months of age [1], with a recurrence rate of 5% in these patients [2]. A congenital skull base malformation is the causes of recurrent meningitis in 43-70% of bacterial meningitis cases among young children [3,4], whereas skull defects secondary to traffic accidents account for 53% of adult cases [2]. However, there were some reports of elderly patients with a spontaneous cerebrospinal fluid (CSF) fistula, which formed over many years [3,4]. In these patients, an association with cystic lesions has been suggested, but the underlying mechanism is not fully understood [1-5]. Further discussion on how to diagnose and evaluate cystic lesions is also required. We report a case of recurrent meningitis in an 11-year-old girl with a cystic lesion complicated by a fistula in the petrous apex and cystic lesion enlargement over 2 years.

Case Report

An 11-year-old Japanese girl was transferred to our hospital with fever, severe headache, and vomiting. Her Rohrer index was 100.7. She was diagnosed with meningitis with CSF findings, and *Streptococcus pneumoniae* was detected in a CSF culture. Her consciousness recovered to normal after antibiotic treatment (ceftriaxone, 100 mg/kg/day on Days 1-14; vancomycin, 40 mg/kg 2 times/day on Days 1-14).

Post-discharge, the patient experienced occasional headaches, and serous fluid was detected several times on the left side of her nose, implying CSF rhinorrhea. She developed meningitis for the second time 8 months after the first episode.

We examined whether the recurrent meningitis was triggered by immunological disorders such as complement defects or immunoglobulin subclass defects; no abnormality was found. She had no history of head

trauma or skull base surgery. Computed tomography (CT) was performed to identify the cause of the meningitis, and a cystic lesion accompanying the osteolytic lesion was detected at the left petrous apex. Another abnormality shown on the CT image was canals in the basiocciput. We diagnosed incomplete canalis basilaris medians (CBM; Fig. 1). We considered that either the cystic lesion or the incomplete CBM had a fistula causing the recurrent meningitis, but we could not detect a fistula on CT images.

On magnetic resonance imaging (MRI), we observed effusion in the petrous apex bone around the cystic lesion, suggesting the existence of a fistula (Fig. 2); there was no indication of a fistula around the CBM. The effusion around the cystic lesion did not resolve after the meningitis was treated, which suggests that the effusion was not an inflammatory reaction. On MRI, the cystic lesion was identical to the CSF, and it seemed hypointense on T1-weighted images and hyperintense on T2-weighted and fluid attenuated inversion recovery (FLAIR) images (Fig. 2); this was suggestive of an arachnoid cyst or cephalocele. We suggested surgical intervention to the patient and her parents but we were unable to obtain consent because of the potential risk of postoperative sequelae such as facial nerve paralysis. We decided to follow up after administering *Haemophilus influenzae* type B and polyvalent 23 pneumococcal vaccines.

After being discharged, the patient often complained of headaches but had no recurrence of meningitis. CT performed 2 years after the second meningitis episode revealed an enlarged osteolytic lesion. The

diameter of the cystic lesion was enlarged 4 mm in axial images and 6 mm in coronal images (Fig. 3). We again suggested surgical intervention, and the patient and her family agreed to the surgery. Surgery for the cystic lesion was performed using the trans-middle cranial fossa approach. A fistula was identified on the posterior side of the cystic lesion, and CSF leaking in accord with the pulsation was also identified. We diagnosed an arachnoid cyst based on the surgical findings of absence of dural lining. The fistula was repaired using tissue from the temporal muscle and its fascia. The patient experienced slightly decreased sensation on the left side of her face after the surgery, but no other sequela were observed.

Discussion

Congenital arachnoid cysts are thought to form at around 15 weeks' gestation, and the petrous apex is the second most common location for arachnoid cysts [6], whereas an acquired arachnoid cyst can result mainly from trauma [7]. In our patient's case, as there was no history of trauma, a congenital arachnoid cyst was suspected. Arachnoid cysts typically become symptomatic before 20 years of age, but few cases of recurrent meningitis have been reported in adults. In these cases, arachnoid cysts occurred with dural defects causing osteolysis and a fistula over a long period [8, 9].

Although the mechanism of fistula formation by cystic lesions is not fully elucidated, Kaufman *et al.* suggested that CSF pressure and pulsations might influence the pneumatic sites of bones and lead to fistula

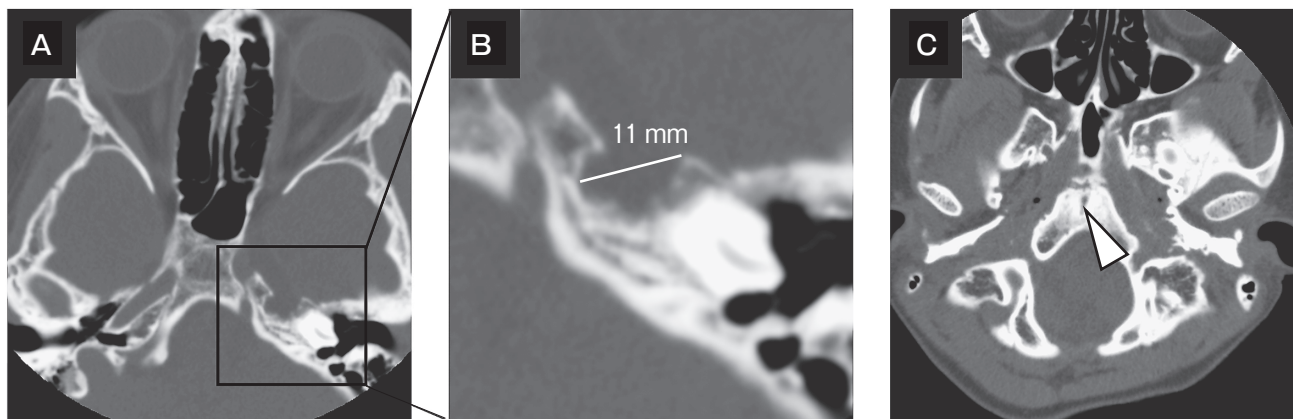


Fig. 1 A, CT showing an osteolytic lesion in the left petrous apex; B, Enlarged image of the osteolytic lesion; C, Canalis basilaris medians (CBM) was also found in the CT, which revealed the incomplete type.

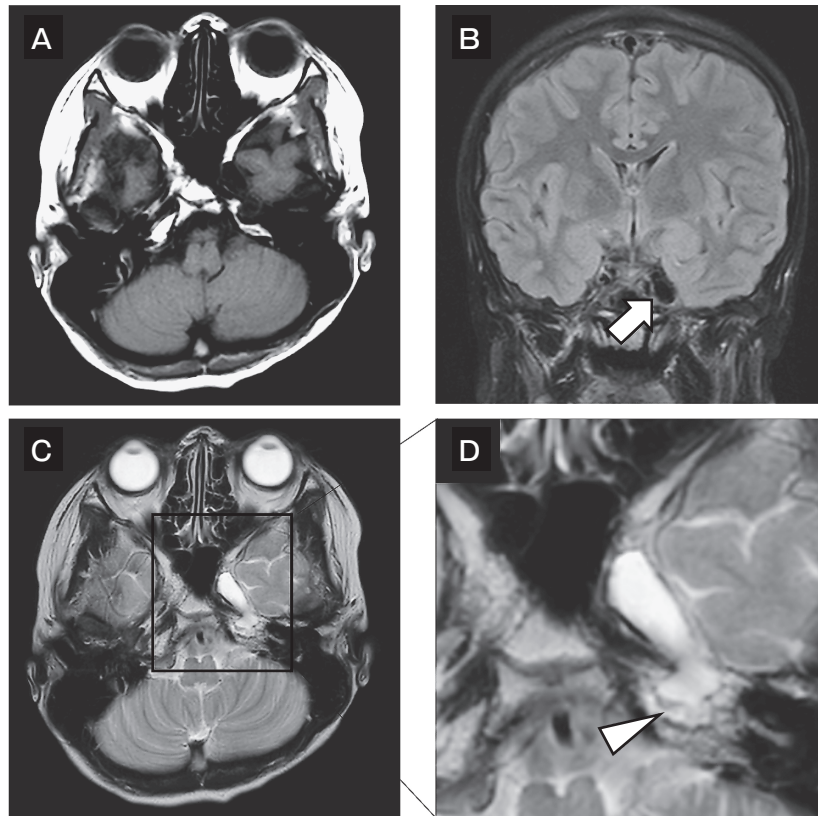


Fig. 2 T1-weighted and FLAIR images showing the cystic lesion as hypointense (A) (arrow in B), and T2-weighted images showing the cystic lesion as hyperintense (C), indicating CSF in the cystic lesion; D, MRI T2-weighted image showing petrous apex cancellous bone as hyperintense identically to the cystic lesion, suggesting that fluid was leaking from the fistula (arrowhead).

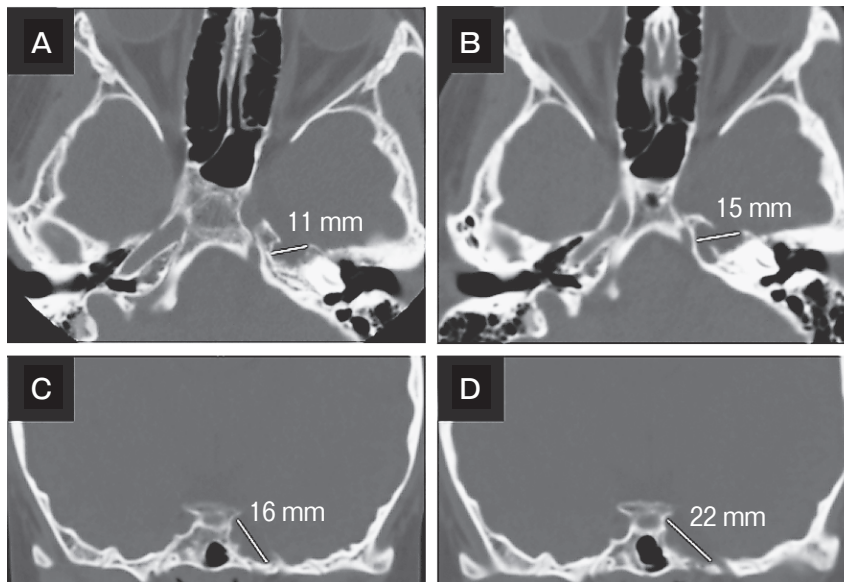


Fig. 3 CT imaging performed on admission of the second meningitis episode (A, C) and 2 years thereafter (B, D). CT imaging performed 2 years after the second meningitis episode showed that the osteolytic lesion had enlarged by 4 mm in the axial image (B) and by 6 mm in the coronal image (D).

formation [5]. In addition to arachnoid cysts and a cephalocele, aberrant large arachnoid granulations (AGs) can also cause a fistula by pulsation. Bone erosion by these lesions were reported mainly in the petrous apex parts of the temporal bones or in the mastoid parts of the temporal bones [2, 10-13], and both bone lesions are anatomically well-pneumatized. Elevated intracranial pressure was also thought to be a central factor in some reports [3, 13]. Females aged 40-60 years with a body mass index >30 tend to show high intracranial pressure and are thought to be at risk of developing a CSF fistula [3, 10, 14].

Our patient had none of the risk factors described above, and she had normal CSF pressure. There is also a report of a patient with a non-traumatic and normal-pressure CSF fistula, which indicating elevated CSF pressure is not essential. Fistulas caused by pulsatility of the basilar artery have also been reported and in these cases, overpneumatization of the clival bone [15], Marfan's syndrome, or repeated Valsalva maneuvers [16, 17] were suspected as factors of a fistula forming in the clivus. Common points in these theories of osteolysis are pulsation, and we speculate that bone erosion by cystic lesions occurs when the factors that strengthen the pulsation or weaken the bone stability coexist.

It is not known how long it takes a fistula to form by an arachnoid cyst; most of the patients with a fistula caused by cystic lesions are adults [4, 5, 13, 14, 18, 19]. Additionally, because most patients with a fistula caused by aberrant AGs were over 50 years old and the size of the aberrant AGs were 2-3 mm [13, 14, 20], it was estimated that a CSF fistula caused by aberrant AGs took several decades to form [11, 14, 20]. A fistula caused by arachnoid cysts was also estimated to form over a long period of time [4]. However, our patient was 11 years old, and the diameter of the osteolytic lesion enlarged by 4 mm on the axial CT slice images and by 6 mm on the coronal CT slice images in the 2 years after she was diagnosed with recurrent meningitis, indicating that a fistula can form faster than expected compared to past case reports.

In our patient, CBM was also observed as another skull base malformation. CBM is an anomaly of the basiocciput, and it consists of well-defined channels. Although there are some cases of repetitive meningitis with CBM completely transversing the basiocciput [21], no case with incomplete CBM and repetitive meningitis has been reported, to our knowledge.

CT or MRI scanning is performed to determine the underlying cause of recurrent meningitis. Recurrent meningitis is often caused by skull anomalies. In this context, CT is helpful because it can provide detailed bone images. Some studies reported that MRI can show a small fistula which cannot be detected by CT [19, 22]. In our patient, effusion was detected in the petrous apex bone around the cystic lesion, indicating a fistula. MRI is also useful for diagnosing a cystic lesion. The CT findings of cholesterol granuloma are the same as those for arachnoid cysts, but these disorders can be differentiated by MRI [23]. Although making a differential diagnosis of arachnoid cysts and cephalocele is not always easy by CT or MRI, distinguishing between these two lesions is not necessarily required because the treatment of these two types lesions causing repetitive meningitis are almost the same. In our patient, we made the final diagnosis of an arachnoid cyst after surgery.

There is no established management for cystic lesions in the petrous apex, but surgery is needed for patients with cystic lesions that are enlarging and causing vertigo, hearing loss, or damage to the facial nerve that causes paralysis, in addition to patients with recurrent meningitis [6, 24]. Conversely, because there are also asymptomatic cystic lesions in the petrous apex that require no treatment [12], the symptoms must be evaluated carefully to determine whether they result from the cystic lesions. Additionally, decisions about the necessity of treatment must be made carefully, because the surgery itself can cause sequelae such as fistula formation or facial nerve paralysis.

In conclusion, pulsations associated with cystic lesions can be an underlying cause of recurrent meningitis in pediatric patients, and bone erosion can progress faster than expected depending on the conditions. MRI is recommended to evaluate cystic lesions, especially to determine the presence of a fistula.

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