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Spontaneous Cerebrospinal Fluid Rhinorrhea Associated With a Far Lateral Temporal Encephalocele

—Case Report—

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Abstract

A 35-year-old female complained of right-sided watery nasal discharge persisting for 2 weeks. Neuroimaging investigations revealed a defect in the lateral side of middle cranial fossa, temporal lobe encephalocele protruding into the lateral extension of the sphenoid sinus, and cerebrospinal fluid (CSF) collection on the right side of the sphenoid sinus. The transcranial approach was performed for resection of the encephalocele and obliteration of the cranial base defect anterolateral to the foramen spinosum with transcranial multilayered closure of the defect using autologous fat, cranial bone graft, and vascularized split temporal muscle. At 1-year follow up, the CSF rhinorrhea had not recurred. Transcranial multilayered closure of the defect is safe and reliable, particularly for large CSF fistula at the far lateral sphenoid sinus.

Key words: cerebrospinal fluid, rhinorrhea, temporal encephalocele, sphenoid sinus, surgical treatment

Introduction

Encephalocele usually involves herniation of frontal lobe tissue through an anterior cranial fossa defect into the ethmoid sinus or nasal cavity. Encephalocele can also result from temporal lobe herniation through a middle fossa defect into the sphenoid sinus. Intrasphenoidal encephaloceles are subdivided by location into medial-perisellar and lateral sphenoid recess types.[10] Perisellar encephaloceles within the sphenoid sinus are considered more common, whereas basal encephaloceles limited to the lateral sphenoid sinus are rare. Spontaneous cerebrospinal fluid (CSF) rhinorrhea or even meningitis is often the first manifestation of a previously existing, occult malformation of the skull base.[4] We present a rare case of spontaneous CSF rhinorrhea caused by a far lateral intrasphenoidal encephalocele.

Case Report

A 35-year-old female presented with a complaint of intermittent right-sided nasal watery discharge persisting for 2 weeks. She had no history of birth trauma or other head trauma, and had undergone no previous surgery procedures. A glucose-oxidase test confirmed that the discharge was CSF. Coronal computed tomography revealed development of the bilateral frontoethmoid sinuses and the right sphenoid sinus. A defect in the lateral side of right middle cranial fossa communicated with the lateral extension of the right sphenoid sinus (Fig. 1A). Magnetic resonance (MR) imaging demonstrated brain tissue herniation into the sphenoid sinus through a skull base defect, and CSF collection on the right side of the sphenoid sinus

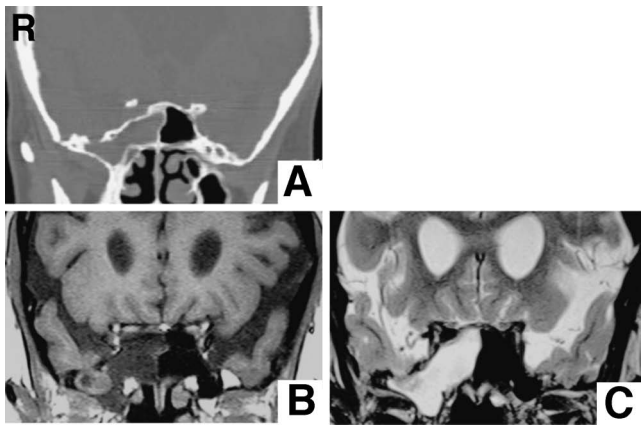

Fig. 1 A: Preoperative coronal bone window computed tomography scan revealing a defect in the far lateral side of right middle cranial fossa and lateral extension of the sphenoid sinus. B: Coronal T_1-weighted magnetic resonance image revealing the temporal lobe herniating into the right sphenoid sinus. C: Coronal T_2-weighted magnetic resonance image revealing cerebrospinal fluid collection in the right sphenoid sinus.

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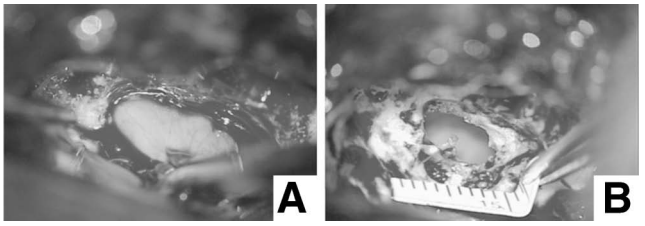

Fig. 2 A: Intraoperative imaging of the temporal encephalocele via the subtemporal extradural approach. B: Intraoperative imaging of the dehiscent dura after resection of the temporal encephalocele.

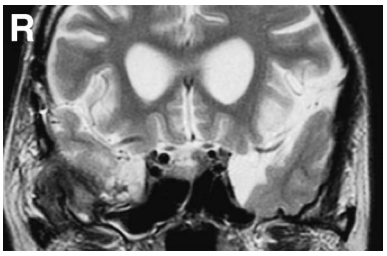

Fig. 3 Coronal T_2-weighted magnetic resonance image obtained 1 month after surgery showing disappearance of the temporal encephalocele and cerebrospinal fluid collection in the right sphenoid sinus.

(Fig. 1B, C), hypogenesis of the lower vermis, and mild ventriculomegaly. No enhanced lesions such as brain tumor or abscess were detected. Intracranial pressure measured by lumbar tap was within the normal range.

Right frontotemporal craniotomy and extradural exploration of the middle cranial fossa was performed. Gliotic encephalocele herniation was observed passing through an about 10-mm round defect anterolateral to the foramen spinosum in the cranial base (Fig. 2A). The encephalocele was resected, revealing the dehiscent dura (Fig. 2B). The dural defect was closed with sutures, then the cranial base defect was filled with multiple layers of autologous fat and cranial bone graft. Furthermore, a vascularized pedicle of the split temporal muscle was laid extradurally in the subtemporal region. Fibrin glue was placed extradurally over the repaired dura. Spinal drainage was maintained for 5 days.

The postoperative course was uneventful. Histological evaluation of the specimens resected by craniotomy showed the typical findings of basal encephalocele, consisting of brain tissue with respiratory mucosa, and no evidence of tumors. The temporal encephalocele and CSF collection in the right sphenoid sinus disappeared on postoperative MR imaging (Fig. 3). The patient had no further leakage during a follow-up period of 1 year.

Discussion

Sphenoid cranial base defects are considered more likely to be of congenital origin than to be acquired. Sternberg's canal is a congenital structure resulting from incomplete fusion of different sphenoid bone components and may act as the site of origin of congenital meningoceles or CSF fistulas.[13] Communication with the middle cranial fossa and CSF fistula is more likely if the sphenoid sinus is laterally pneumatized.[14] Most cases of lateral sphenoid recess type involve a defect of the middle cranial fossa located lateral to the foramen ovale or the foramen rotundum.[4,6,10,11] In our present case, there was a bony defect anterolateral to the foramen spinosum, and was thought to be the far lateral sphenoid recess type. CSF pressures and the hydrostatic pulsative forces may lead to the development of pit holes on the middle fossa at the sites of arachnoid villi with herniation of dura/arachnoid or brain tissue.[7] If such defects are located over the underlying lateral extension of the sphenoid sinus, encephalocele can develop and lead to CSF leakage into the sinus. In previous interesting cases, intracranial pressure was raised after the closure of the fistula and ventriculoperitoneal shunting was required.[3,9] Ventriculoperitoneal shunt operation may be necessary if CSF production increases due to the chronic rhinorrhea,[3] or if the intracranial pressure is high.[9] Our patient showed normal intracranial pressure before surgery and did not develop hydrocephalus after repair of the CSF fistula.

Temporal lobe encephalocele protruding through a middle fossa defect is rare with few distinctive features, so is difficult to identify. The mean delay between onset of symptoms such as CSF rhinorrhea and the diagnosis was 13.1 months in 15 patients.[4] Without repair of the defect, patients with CSF leakage have increased risk of developing ascending intracranial complications such as meningitis, brain abscess, or seizure.[4,10]

Historically, transcranial surgery has been offered for fistula involving the lateral extension of the sphenoid sinus,[2,5,6,8,11,15] but recently endoscopic approaches have been successfully performed for the repair of CSF leakage and encephaloceles.[1,4,10,12] Fistulas involving lateral extension of the sphenoid sinus require a transcranial approach for direct visualization and obliteration of the defect, whereas fistulas involving the central portion of the sinus may be successfully obliterated transsphenoidally, as endoscopic transnasal repair of fistulas involving lateral extension of the sphenoid sinus had failed in three of the four patients.[11] Intracranial repair with a vascularized calvarial graft is an excellent method to close large bony defects in the cranial base.[5,15] If the size of the encephalocele and its bony defect is greater than 1 cm, an intracranial surgical approach to the middle cranial fossa is required for multilayered closure to prevent complications.[2] In our present case, although the cranial base defect was large, transcranial multilayered closure of the defect using autologous fat, cranial bone graft, and vascularized split temporal muscle prevented recurrence of CSF leakage.

The present case of spontaneous CSF rhinorrhea caused by far lateral intrasphenoidal encephalocele suggests that early diagnosis of spontaneous CSF rhinorrhea and surgical treatment is essential to prevent severe complications. Although less invasive endoscopic surgery has been suc-

cessfully performed for the repair of CSF leakage recently, transcranial multilayered closure of the defect is safe and reliable, particularly for large CSF fistula at the far lateral sphenoid sinus.

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