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Case Report

Lateral Craniopharyngeal Canal: An Anatomical Cause of Spontaneous Rhinorrhea

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ABSTRACT

The aim of this report is to investigate the pathogenesis and surgical treatment of encephalocele located in the sphenoid sinus and presented with spontaneous rhinorrhea. The data of a patient with sphenoid sinus encephalocele was analyzed in association with the review of literature. The patient admitted to our clinic with rhinorrhea from the left nasal cavity. Cranial magnetic resonance imaging revealed bone defects in the left sphenoid sinus wall, and the presence of brain tissue and cerebrospinal fluid (CSF) in the sphenoid sinus. The patient underwent dural repair via left pterional approach. Rhinorrhea disappeared after surgery. The pathogenesis of the encephalocele in the sphenoid sinus is not clear. It may be related to the presence of lateral craniopharyngeal canal, extra-gasification in the sphenoid sinus, high intracranial pressure or other factors. It is very important to diagnose accurately the source of CSF fistula in preoperative period, choose the specific operation technique, and follow-up the patient for a long time period.

KEYWORDS: Sphenoid sinus encephalocele, Spontaneous rhinorrhea, Lateral craniopharyngeal canal

INTRODUCTION

Rhinorrhea is common after trauma or operations. Spontaneous rhinorrhea is very rare. Cerebrospinal fluid (CSF) fistula (rhinorrhea) requires 2 anatomical conditions; first is the dural tear and the second is anatomical link with a sinus or nasal cavity.

Lateral craniopharyngeal canal (Sternberg's canal) is a concept proposed by Maximilian Sternberg in 1888, which refers to the sphenoid dysplasia, appeared in the embryonic fissure (3,6,12). Lateral craniopharyngeal canal is only covered by a weak connective tissue. Its incidence is low (about 1/35,000). This canal is an important anatomical factor on the protrusion of brain tissue into the sphenoid sinus through the sphenoid sinus wall (3). Spontaneous CSF leakage may easily develop through the dural defect and lateral craniopharyngeal canal because it is a narrow passage and repeated impacts of CSF facilitate protrusion of the brain tissue into the sphenoid sinus. Surgery is the only effective treatment method of spontaneous rhinorrhea. Transcranial approach and endoscopic transnasal approach are the surgical options. In recent years, a small amount of similar cases were reported in the literature (4,10,11).

We report a patient with spontaneous rhinorrhea who had a lateral craniopharyngeal canal and the brain tissue protruded into the sphenoid sinus through this canal. The patient underwent surgical treatment via pterional craniotomy and recovered well after a follow-up period of 15 months.

CASE REPORT

A 58 year-old male patient admitted to our clinic for a 6 month history of "repeated fluid drainage from the left nasal cavity". The fluid was clear, aggravating as bow or bent, without headache and fever. His neurological examination was normal.

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After admission, the patient underwent lumbar puncture, and the pressure was 150 mm H_2O. Cranial magnetic resonance imaging (MRI) showed communication between the sphenoid sinus and intracranial space, and a bone defect, with a diameter of about 5 mm in the left sphenoid sinus wall. There was also CSF signal intensity and soft tissue in sphenoid sinus. Soft tissue was consistent to the brain parenchyma corresponding with a lateral craniopharyngeal canal (Figure 1A, B). Three days later, the patient underwent left pterional approach to repair the dural defect on the skull base. During the surgery, it was seen that the medial temporal lobe protruded into the sphenoid sinus and regional dura mater contained more than 10 oval holes, like fishnet, ranging about 3-6 mm in diameter (Figure 2A, B). The dural defect was repaired by muscle tissue and collagen sponge, and sealed by fibrin glue. The patient underwent repeated postoperative lumbar punctures. The CSF pressure was between 150 and 180 mm H_2O. There was no other lesion in the postoperative computed tomography (CT) (Figure 3A, B). After follow-up for 15 months, the patient had no rhinorrhea.

This study was conducted in accordance with the declaration of Helsinki with the approval from the Ethics Committee of General Hospital of Fuzhou Military Area Command Nanjing. Written informed consent was obtained from the patient.

DISCUSSION

The orifice of a CSF fistula is usually located in the ethmoid sinus wall, but it may be also seen in the sphenoid sinus wall. Lai et al. (8) reported an incidence of encephalocele or meningocele in the skull base about 1/35,000.

Lateral craniopharyngeal canal is a concept proposed by Maximilian Sternberg in 1888, which is a sphenoid sinus dysplasia appeared in the embryonic fissure. Sphenoid sinus was formed in the third month of embryonic period, ossificated by a plurality of ossification center, which is mainly divided into five parts: the front part of the sphenoid sinus, the lesser wing, sphenoid body, bilateral pterygoid process, back of sphenoid body, greater wing of the sphenoid bone. Each part respectively merges after ossification, except the

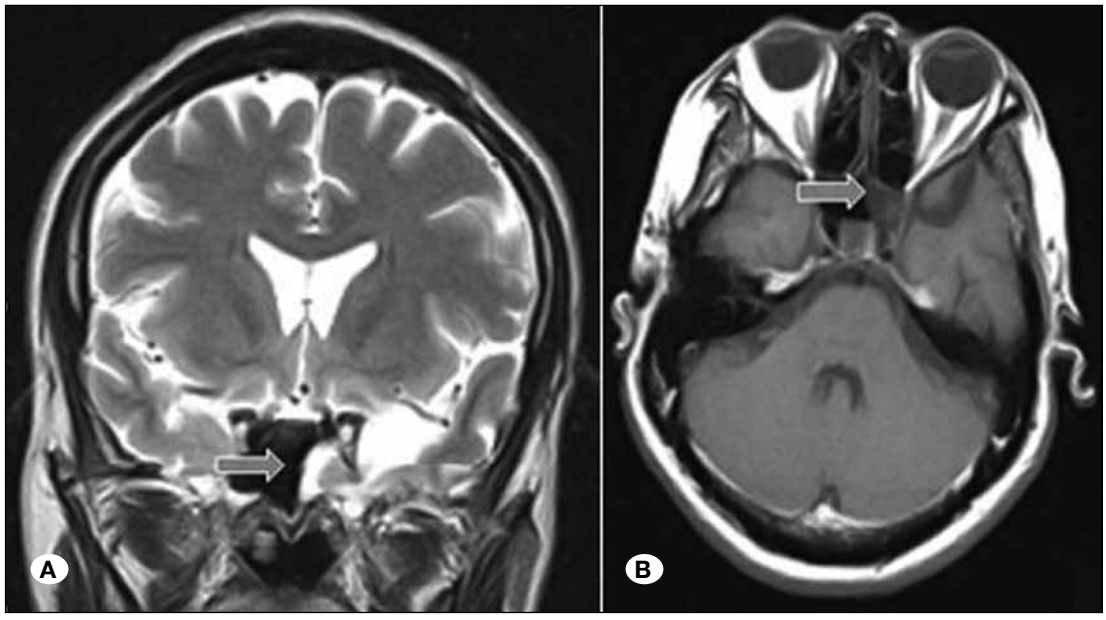

Figure 1: Preoperative cranial MRI scans **(A)** T2 coronal and **(B)** T1 axial slices. The arrow shows Sternberg 's canal, brain tissue protruding into the sphenoid sinus, and accompanied by CSF signal, about the size of 1.3 x 1.4 cm.

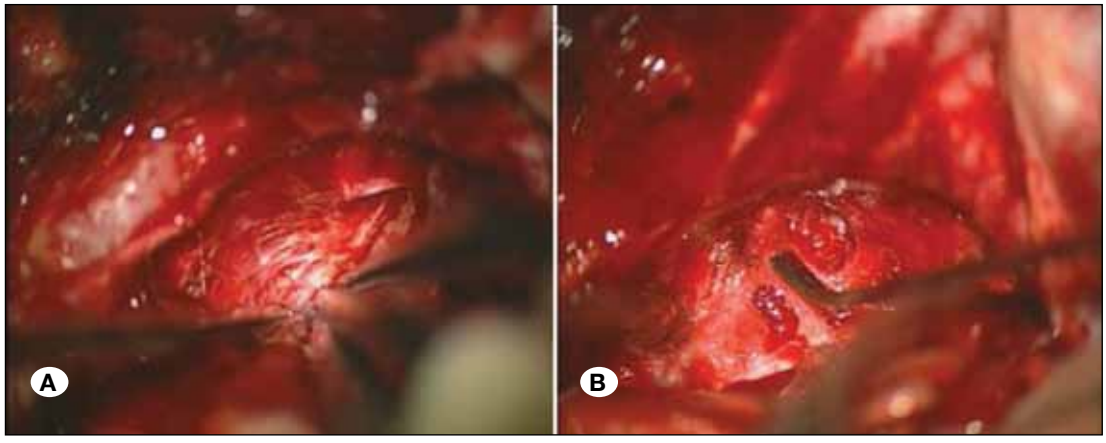

Figure 2: Intraoperative view **(A** and **B)** of the lateral craniopharyngeal canal shows epidural reticulation, maximum diameter of 6 mm.

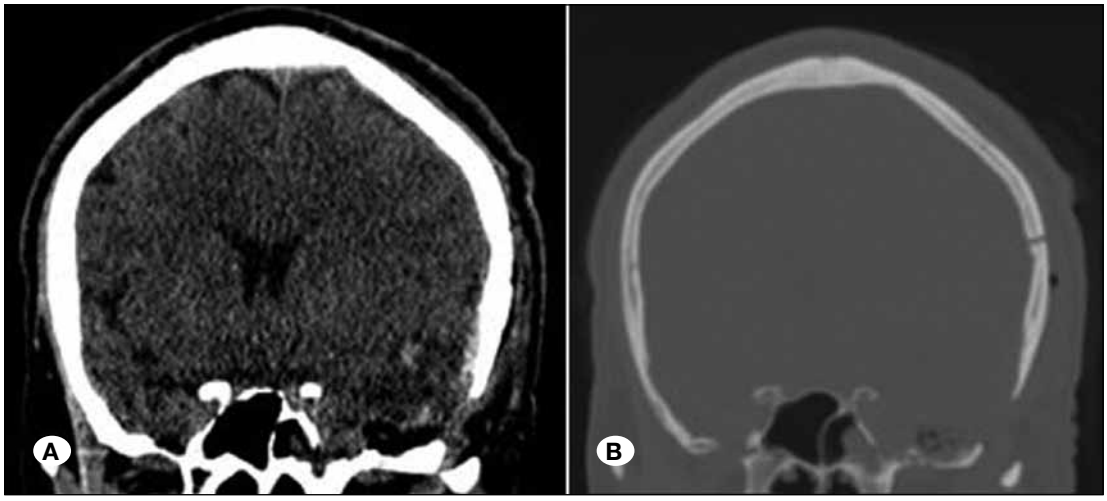

Figure 3: Cranial coronal CT scans **(A** and **B)** one day after operation.

rear part of the greater wing of sphenoid bone. The rest has completely fused at birth or after the birth. When one side of the greater wing of the sphenoid bone and body of sphenoid bone not fused completely, the gap between these parts of the sphenoid bone forms lateral craniopharyngeal canal (4). When Sternberg described the lateral craniopharyngeal canal, he also estimated an incidence about 4% in adults. But in the series of Baranano et al. (3), only one patient had lateral craniopharyngeal canal among the CT scans of 1000 normal volunteers. This canal is covered only by a connective tissue which is relatively weak. It is an important anatomical passage on the sphenoid sinus wall for the protrusion of brain tissue into the sphenoid sinus. Schick et al. (12) and Blaivie et al. (5) suggested that Sternberg's canal is the sign to distinguish the congenital and acquired sphenoid sinus encephalocele. In our patient, Sternberg's canal was identified on the MRI and CT scans as communicating with the front of left sphenoid sinus wall.

In the previous imaging studies on the spontaneous CSF leakage, many scholars did not find this kind of patient who had sphenoid sinus encephalocele and Sternberg's canal. But, it should be paid attention to the relationship of CSF fistula with the primary empty sella. Shetty et al. (15) reported that 7 of 11 patients with spontaneous CSF leakage had also empty sella. Schlosser and Bolger (13) investigated the images of 15 patients with spontaneous CSF leakage. Ten of them had completely empty sella and other 5 had partial empty sella. In the meantime, among the 9 patients with non-spontaneous CSF leakage, only one had partial empty sella.

Empty sella has been widely considered as an indirect sign of increased intracranial pressure, suggesting the correlation between the intracranial pressure and the spontaneous CSF leakage. Schlosser et al. (14) measured CSF pressure following the dural repair in 8 cases with spontaneous CSF leakage. They found that the CSF pressure in 7 cases was higher than the normal pressure (average 325 mm H_2O), and 3 patients with traumatic CSF leakage served as control group (average 140 mm H_2O) in this study. High intracranial pressure increases the CSF pressure on the surrounding meningeal tissue and has a beating effect on the surrounding structures. When there is a local bone defect around this dura mater, it is easy to form or enlarge a dural hole and cause spontaneous CSF leakage.

Under normal circumstances, bony structures around the sphenoid sinus wall can well withstand the CSF pressure. When the intracranial pressure increased (secondary to obesity or hydrocephalus), or bone bearing capacity decreased (due to lateral craniopharyngeal canal or osteoporosis), the sphenoid sinus encephalocele and spontaneous rhinorrhea may develop. Our patient has normal preoperative intracranial pressure, and MRI shows no empty sella, brain herniation in front of the sphenoid sinus wall in association with lateral craniopharyngeal canal. At the same time, the intraoperative reticulation damage of dura is visible, which meets the criteria of rhinorrhea. So, we considered that the spontaneous rhinorrhea in our patient was started by the formation of lateral craniopharyngeal canal.

Spontaneous rhinorrhea has specific clinical symptoms, so the clinical diagnosis is not difficult. High-resolution CT scan with thin slices can display the existence of Sternberg's canal, and MRI can well show the CSF and brain tissue signal in the sphenoid sinus. There are the clinical manifestations of rhinorrhea in one side, and the fistula may locate in the contralateral side or bilateral due to separation in sphenoid sinus (15). So, preoperative accurate determination of fistula site is very important for the selection of best surgical approach. Some scholars performed radiography with subarachnoid contrast agent injection, which provides greater accuracy on the detection of fistula.

There is no report on the self-healing of lateral craniopharyngeal canal combined with CSF leakage. Operation is currently the only treatment method for the repair of fistula. Pterional approach is usually the first choice. In our patient, we used the traditional pterional approach to repair the fistula, and there was no recurrence after operation. This approach can clearly expose the anterior and middle skull base structures, reveal the fistula, with broad vision. Many authors also used this technique and achieved satisfactory results (1, 7). With minimally invasive concept, the endoscopic technique is

becoming more popular, and many surgeons preferred endoscopic technique to repair the skull base CSF leakage. Landreneau et al. (9) used transsphenoidal endoscopic approach to repair the CSF fistula. For the fistula located in the middle of sphenoid sinus, the authors reported satisfactory results, but for the fistula in the lateral walls of sphenoid sinus, they reported only 25% of success. For the fistula located in the lateral wall, some authors used the lateral pterygoid process -the maxillary sinus- sphenoid sinus approach and the pterygoid process -Sieve transsphenoidal approach (2,6). These two approaches can better expose the lateral wall of sphenoid sinus than the other approaches.

We should choose operation technique and approach according to the fistula position and the habits of the surgeon. After the surgical repair, the CSF leakage may disappear and intracranial pressure may increase. This is one of the reasons of recurrence in rhinorrhea. Therefore, long-term follow-up is necessary.

CONCLUSION

Spontaneous CSF leakage is a rare clinical condition. Its pathogenesis is not clear. The presence of lateral craniopharyngeal canal causes the brain tissue herniation into the sphenoid sinus, which is one of the anatomical bases of spontaneous rhinorrhea. Diagnosis is not difficult with CT and MRI scans. Accurate positioning is very important for the treatment of fistula. Operation technique and approach should be individualized and long-term follow-up is important after surgery. Special attention should be paid to the occurence of intracranial hypertension.

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