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Case Report

Cerebrospinal fluid rhinorrhea with meningoencephalocele related to Sternberg's canal: A report of two cases

Satoshi Adachi[1], Hideaki Ueno[2], Shunsuke Magami[1], Naohide Fujita[1], Shintaro Nakajima[1], Ryogo Ikemura[1], Yasuhito Ueki[1], Yuki Takaki[1], Keisuke Murofushi[1], Yasuaki Nakao[1], Takuji Yamamoto[1]

[1]Department of Neurosurgery, Juntendo University Shizuoka Hospital, Izunokuni, [2]Department of Neurosurgery, Juntendo University Urayasu Hospital, Urayasu, Japan.

E-mail: *Satoshi Adachi - s-adachi@juntendo.ac.jp; Hideaki Ueno - hideakiueno1229@gmail.com; Shunsuke Magami - smagami@juntendo.ac.jp; Naohide Fujita - n-fujita@juntendo.ac.jp; Shintaro Nakajima - snnakaji@juntendo.ac.jp; Ryogo Ikemura - r-ikemura@juntendo.ac.jp; Yasuhito Ueki - y-ueki@juntendo.ac.jp; Yuki Takaki - y.takaki.kf@juntendo.ac.jp; Keisuke Murofushi - murofushi7735@gmail.com; Yasuaki Nakao - y-nakao@juntendo.ac.jp; Takuji Yamamoto - tyamamoto@med-juntendo.jp

***Corresponding author:**
Satoshi Adachi,
Department of Neurosurgery,
Juntendo University Shizuoka Hospital, Izunokuni, Japan.

s-adachi@juntendo.ac.jp

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ABSTRACT

Background: Cerebrospinal fluid (CSF) rhinorrhea with meningoencephalocele (MEC) associated with Sternberg's canal is rare. We treated two such cases.

Case Description: A 41-year-old man and a 35-year-old woman presented with CSF rhinorrhea and mild headache worsening with standing posture. Head computed tomography showed a defect close to the foramen rotundum in the lateral wall of the left sphenoid sinus in both cases. Head magnetic resonance (MR) imaging and MR cisternography revealed that brain parenchyma had herniated into the lateral sphenoid sinus through the defect of the middle cranial fossa. The intradural and extradural spaces and bone defect were sealed with fascia and fat through both intradural and extradural approaches. The MEC was cut away to prevent infection. CSF rhinorrhea completely stopped after the surgery.

Conclusion: Our cases were characterized by empty sella, thinning of the dorsum sellae, and large arteriovenous malformations that suggest chronic intracranial hypertension. The possibility of Sternberg's canal in patients with CSF rhinorrhea with chronic intracranial hypertension should be considered. The cranial approach has the advantages of lower infection risk and the ability to close the defect with multilayer plasty under direct vision. The transcranial approach is still safe if performed by a skillful neurosurgeon.

Keywords: Arteriovenous malformation, Cerebrospinal fluid rhinorrhea, Lateral craniopharyngeal canal, Meningoencephalocele, Surgical repair

INTRODUCTION

Sternberg's canal (lateral craniopharyngeal canal) was first described by Maximilian Sternberg in 1888,[14] and is considered to be an underlying factor in the occurrence of sphenoid sinus spontaneous cerebrospinal fluid (CSF) rhinorrhea. However, the incidence of CSF rhinorrhea with meningoencephalocele (MEC) caused by the canal is low.[13] We describe our experience with two more cases.

CASE REPORTS

Case 1

A 41-year-old man became aware of left-sided intermittent nasal discharge which gradually worsened and mild headache worsening with standing posture. He had no previous history of head trauma, medical disorders, or seizures. Clinical examination revealed left-sided CSF rhinorrhea, and biochemical tests showed protein 46 mg/dL and glucose 75 mg/dL in the nasal discharge. The rhinorrhea worsened with cervical anteflexion. Head computed tomography (CT) showed a defect lateral to the foramen rotundum in the lateral wall of the left sphenoid sinus and erosion of the dorsum sellae [Figures 1a and b]. Head magnetic resonance (MR) imaging and MR cisternogram revealed brain parenchyma had herniated into the lateral sphenoid sinus through the defect of the middle cranial fossa and the empty sella [Figures 1c-e]. Surgical treatment was performed to repair the defect of the skull base following lumbar drainage. Left frontotemporal craniotomy and temporal extension were performed. Intraoperatively, the fragile dura mater and the dural defect were detected on the middle cranial fossa. Small amounts of brain tissue including the arachnoid membrane of the temporal lobe had protruded through the dura mater. The epidural procedure showed that the protruded brain tissue was blocking the bony defect leading to the sphenoid sinus. The herniated MEC was cut away followed by suturing of the dural defect. The intradural and extradural spaces and bone defect were sealed with abdominal external oblique fascia containing fat, fibrin glue, and biological tissue reinforcement material through both the intradural and extradural approaches [Figures 2a-g]. Histopathological examination of the tissue sample confirmed herniation of gliotic brain parenchyma and meningeal tissue. The CSF rhinorrhea completely stopped after the surgery. He was discharged without rhinorrhea and headache at 7 days after surgery. The CSF rhinorrhea has not recurred for over 1 year.

Case 2

A 35-year-old woman suffered intermittent nasal discharge without seizures. She had a history of large arteriovenous malformations (AVMs) in the right frontal lobe. CT showed a defect in the floor of the sella turcica and AVMs in the right frontal lobe, with nidus of 5 cm [Figures 3a-c]. MR cisternogram revealed the empty sella with intracranial and abnormal fluid collection in the sphenoid sinus [Figure 3d]. The defect in the floor of the sella turcica was repaired through the transsphenoidal approach followed by lumbar peritoneal shunt, resulting in cessation of the CSF rhinorrhea. She again became aware of nasal discharge at 1 year after the first surgery. Head CT showed severe pneumocephalus and a defect between the foramen rotundum in the lateral wall of the left sphenoid sinus and the foramen ovale, and erosion of the dorsum sellae and many ovoid bony defects in the middle cranial fossa [Figures 4a and b]. Head MR cisternogram revealed that brain parenchyma had herniated into the lateral sphenoid sinus through the defect of the middle cranial fossa [Figures 4c and d]. Left frontotemporal craniotomy with temporal extension revealed the middle cranial dura involved multiple holes and protruded with brain parenchyma into the skull base defect between the foramen rotundum and foramen ovale in the middle skull base leading to the sphenoid sinus. The herniated MEC was cut away and the nasal mucosa was exposed. The bone defect was reconstructed with a titanium plate. The intradural and extradural spaces including the bone defect were sealed with temporal fascia, abdominal fat, fibrin glue, and biological tissue reinforcement material [Figures 5a-h]. The CSF rhinorrhea completely stopped after the surgery.

DISCUSSION

Various classifications of CSF rhinorrhea have been reported. CSF rhinorrhea was divided into three categories as follows: (1) traumatic, (2) postsurgical, and (3) spontaneous.[18] CSF rhinorrhea can also be divided into traumatic and nontraumatic categories.[9] Nontraumatic CSF rhinorrhea was grouped

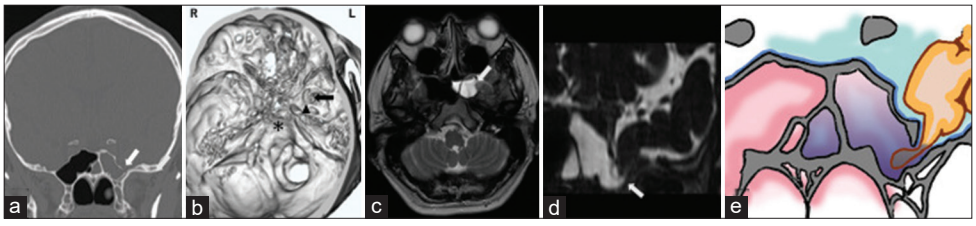

Figure 1: Case 1 - (a and b) Computed tomography scans showed a defect (white arrow, black arrow) in the lateral wall of the left sphenoid sinus lateral to the foramen rotundum (black arrowhead) and erosion of the dorsum sellae (asterisk). (c and d) Head T2-weighted magnetic resonance (MR) image (c) and MR cisternogram (d) revealed brain parenchyma had herniated into the lateral sphenoid sinus through the defect of the middle cranial fossa (white arrow). (e) Schema of meningoencephalocele and cerebrospinal fluid rhinorrhea associated with Sternberg's canal.

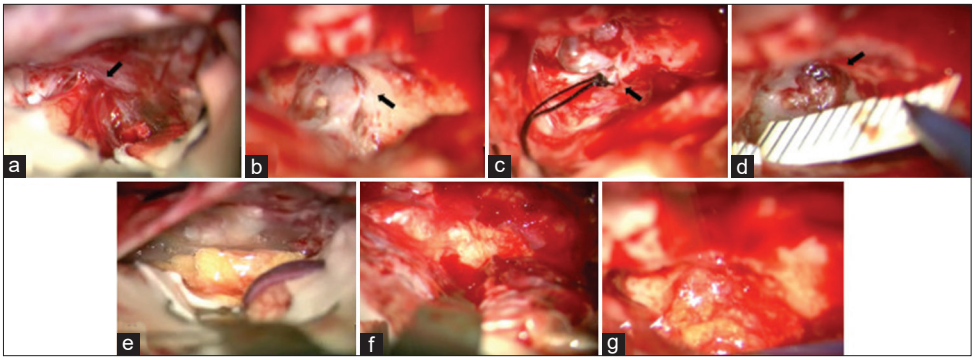

Figure 2: Case 1 - Intraoperative photographs showing the left frontotemporal craniotomy with temporal extension. (a) The intradural approach exposing middle cranial dura and brain tissue protruding into the defect in the middle skull base leading to the sphenoid sinus (arrow). (b) Herniated dura (arrow). (c) Dural defect was sutured through the extradural approach (arrow). (d) Defect in the lateral sphenoid sinus was 5 mm in size (arrow). (e-g) Intradural (e) and extradural spaces (f) and bone defect (g) were sealed with abdominal external oblique fascia containing fat, fibrin glue, and biological tissue reinforcement material.

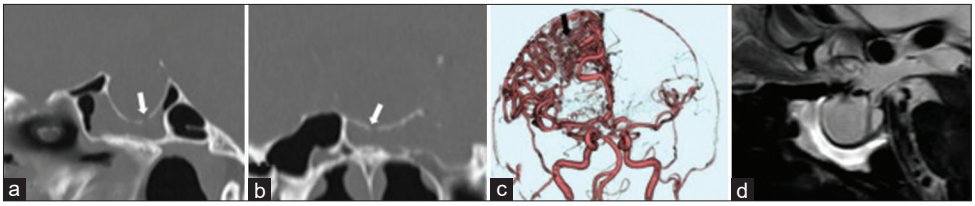

Figure 3: Case 2 - (a and b) Sagittal (a) and coronal computed tomography scans (b) showed a defect in the floor of the sella turcica (arrow). (c) Arteriovenous malformations in the right frontal lobe, with nidus of 5 cm (arrow). (d) Magnetic resonance cisternogram revealed abnormal fluid collection in the sphenoid sinus and the empty sella.

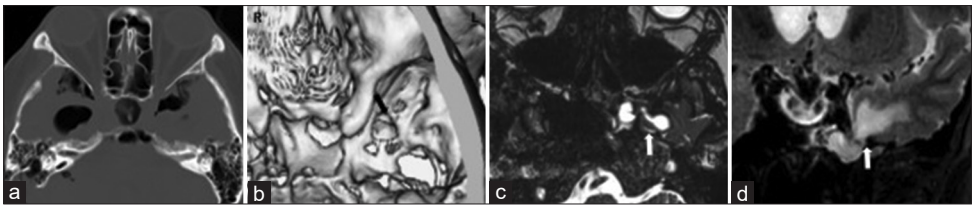

Figure 4: Case 2 - (a and b) Computed tomography scans showed severe pneumocephalus and a defect in the lateral wall of the left sphenoid sinus between the foramen rotundum and foramen ovale (black arrow). (c and d) Axial (c) and coronal (d) magnetic resonance cisternogram showed that brain parenchyma had herniated into the lateral sphenoid sinus through the defect of the middle cranial fossa (white arrow).

into two major subdivisions as follows: (1) normal pressure leaks with congenital anomalies, osteomyelitic erosion, and focal atrophy which includes olfactory and intrasellar (empty sella), and (2) high pressure leaks with tumors or hydrocephalus. Here, we presented cases of rhinorrhea due to a defect in the sellar floor with the empty sella acting as a tense pulsating cyst. MR imaging showed the sellar floor was intact despite the presence of empty sella, but brain parenchyma had herniated into the lateral sphenoid sinus through a defect of the middle cranial fossa in our Case 1. Sphenoid sinus is a potential origin for spontaneous CSF rhinorrhea.

Sternberg's canal (lateral craniopharyngeal canal) has been considered to be an underlying factor related to sphenoid sinus spontaneous CSF rhinorrhea.[4,13] The incidence of persistent patent Sternberg's canal ranges from 0.42% to 6.1% in adults.[17] Sternberg's canal may persist due to ossification disturbances within the fusion planes of the sphenoid sinus in childhood. Before final fusion, a small canal connecting the middle cranial fossa with the nasopharynx is created, called

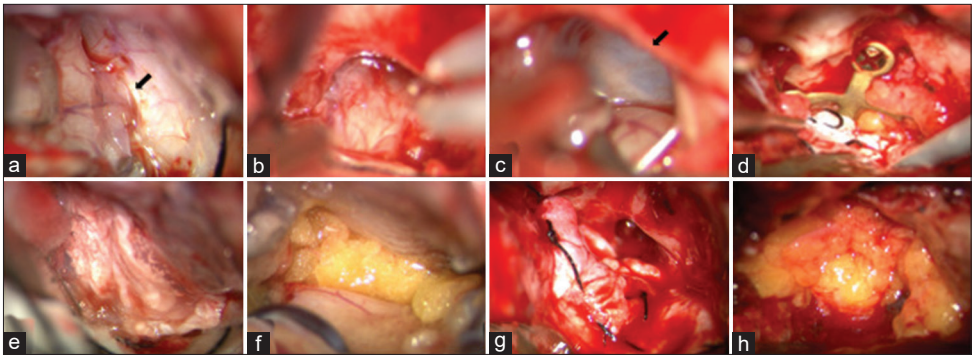

Figure 5: Case 2 - Intraoperative photographs of the second surgery. (a and b) Intradural (a) and extradural approaches (b) to the Stenberg's canal. The cranial dura involved multiple holes and protruded with brain parenchyma (black arrow) into the skull base defect between the foramen rotundum and foramen ovale in the middle skull base leading to the sphenoid sinus. (c) Herniated dura and meningoencephalocele were cut away and the nasal mucosa (black arrow) exposed. (d) Bone defect was reconstructed with a titanium plate. (e-h) Intradural (e and f) and extradural spaces (g and h) including the bone defect were sealed with temporal fascia, abdominal fat, fibrin glue, and biological tissue reinforcement material.

the lateral craniopharyngeal canal. If the sphenoid sinus may develop to reach the fusion plane, the middle fossa, and the sphenoid sinus, and become a potential source of CSF leak and MEC.[13,17] Various variations have been reported on the position of Sternberg's canal, sometimes medial or lateral to the foramen rotundum, or parasellar lesion.[2,17] Sternberg's canal must exist medial to the superior orbital fissure, the foramen rotundum, and the vidian canal.[4] Therefore, the vast majority of lateral sphenoid CSF leaks do not arise from a patent Sternberg's canal, but congenital weakness may be present caused by the prior existence of Sternberg's canal. Further study of the relationship between Sternberg's canal and MEC is needed.

Rhinorrhea and MEC associated with Sternberg's canal are rare.[1-3,5-8,10-12,15-17] Most cases occurred in adults, and some presented with headache and meningitis. Many cases were characterized by empty sella and arachnoid villi, in female, middle-aged and obese patients with benign intracranial hypertension. These clinical features may affect the already weak structure of the cranium.[2] Our Case 1 showed erosion of the dorsum sellae and the empty sella that indicates chronic intracranial hypertension. Case 2 had large AVMs and empty sella due to chronic intracranial hypertension. The previous rhinorrhea might have resulted from a defect in the sellar floor with the empty sella acting as a tense pulsating cyst. The present rhinorrhea was considered to result from the patent Sternberg's canal and chronic intracranial hypertension. The clinical course seen in our Case 2 was complicated and extremely rare.

Treatment of MEC and CSF rhinorrhea consists of reconstruction through the transcranial or endoscopic approach. Herniated dura mater and brain parenchyma should be excised because these tissues are considered contaminated and functionless.[2] The transcranial approach has the advantages of lower infection risk[1] and the ability to close the defect with multilayer plasty under direct vision, but this approach is highly invasive and carries the risk of epilepsy after surgery. The endoscopic approach may be less invasive with smaller skin incision and easier access to midline lesion in the sphenoid sinus.[2,17] Disadvantages of the endoscopic approach involve difficulty in the approach to the lateral site of the sphenoid sinus and the potential risk of persistent CSF leak and meningitis. The trans-ethmoidal-sphenoidal approach and the trans-ethmoidal-sphenoidal-pterygoidal approach can reach the lateral recess in the sphenoid sinus, but these approaches are sometimes challenging and not actually minimally invasive.[1,2] Recommendation of the optimum treatment is difficult. The transcranial approach is still safe if performed by a skillful neurosurgeon. Surgical method should be decided depending on the operator's expertise and patient's conditions such as the location and size of the leakage point.

CONCLUSION

Our cases were characterized by empty sella and thinning of the dorsum sellae which suggest chronic intracranial hypertension. Furthermore, our Case 2 had a long history of large AVMs and previous CSF rhinorrhea due to a defect in the floor of the sella turcica. The possibility of Sternberg's canal in patients with spontaneous CSF rhinorrhea with chronic intracranial hypertension should be considered. The transcranial closure technique which requires multi-layered reconstruction might be safe and effective if performed by a skillful neurosurgeon. Intracranial pressure management and prolonged clinical follow-up are essential in patients with chronic intracranial hypertension.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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