Fallopian Canal Meningocele with Spontaneous Cerebrospinal Fluid Otorrhea: Case Report and Systematic Review of the Literature

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■ OBJECTIVE: **To present a case of spontaneous cerebrospinal fluid (CSF) otorrhea from a fallopian canal meningocele involving the geniculate fossa and review all cases of fallopian canal CSF leak reported in the literature with discussion of management and outcomes.**

■ METHODS: **A 53-year-old woman with history of morbid obesity and hypertension presented to a tertiary care referral center with unilateral high-volume CSF otorrhea. High-resolution temporal bone computed tomography demonstrated significant dilatation of the geniculate fossa. Rates of postoperative facial paralysis and refractory CSF leak were reported for the present case and prior cases reported in the literature.**

■ RESULTS: **Locations of fallopian canal dehiscence, surgical approaches, techniques for packing dehiscence, rates of postoperative facial paralysis and CSF leak, and revision procedures were reported for the present case and 14 cases in the literature. The present case involved dehiscence of the geniculate fossa that was approached via combined transmastoid–middle cranial fossa exploration with facial nerve monitoring. The area of dehiscence was carefully packed with temporalis fascia, muscle, and artificial dural substitute overlay to repair the CSF leak without injuring the facial nerve. Postoperatively, no facial weakness was noted; however, right-sided high-volume CSF otorrhea persisted. After discussing treatment options, the patient underwent subtotal petrosectomy and blind-sac closure of the external auditory canal the following day. This successfully resolved the CSF leak without causing facial nerve weakness.**

■ CONCLUSIONS: **Fallopian canal meningocele is an exceedingly rare cause of CSF otorrhea. Successful repair requires precise packing of the dilated facial canal to occlude the leak without injuring the facial nerve. For refractory CSF leak, subtotal petrosectomy and closure of the external auditory canal warrants consideration.**

INTRODUCTION

Spontaneous cerebrospinal fluid (CSF) leaks involving the fallopian canal are extremely rare, with only 14 cases reported in the literature.[1–11] This uncommon cause of CSF otorrhea is often difficult to manage given the intimate relationship the meningocele has with the intratemporal facial nerve. We present a case of spontaneous CSF otorrhea from a fallopian canal meningocele involving the geniculate fossa. We also review all other cases of fallopian canal CSF leaks reported in the literature and discuss clinical presentation, diagnosis, etiology, and treatment.

MATERIALS AND METHODS

A clinical chart review was conducted to report a case of spontaneous fallopian canal meningocele with CSF leak managed at the authors' center. Pertinent data regarding clinical presentation, management strategy, and outcome are reviewed. In addition, a comprehensive review of the literature was performed to identify and summarize all previously reported cases. Two electronic databases, PubMed and EMBASE, were searched. Strategies for searching electronic databases were as follows: All Fields, Contains "facial nerve" AND All Fields, Contains "CSF"; All Fields, Contains "fallopian" AND All Fields, Contains "CSF"; All Fields,

Key words
- Cerebrospinal fluid otorrhea
- CSF
- Facial canal
- Facial paralysis
- Fallopian canal
- Meningitis
- Meningocele

Abbreviations and Acronyms
CSF: Cerebrospinal fluid
IIH: Idiopathic intracranial hypertension

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Contains "facial nerve" AND All Fields, Contains "meningocele"; All Fields, Contains "geniculate" AND All Fields, Contains "CSF." We supplemented searches of electronic databases with manual screening of the bibliographies of all retrieved publications. We also searched the bibliographies of recent systematic reviews and other review articles for relevant citations.

RESULTS

Case Report

A 53-year-old woman with history of morbid obesity (body mass index 59 kg/m^2) and hypertension presented with mild right-sided conductive hearing loss and high-volume clear otorrhea in the setting of recent myringotomy for suspected serous otitis media. Fluid testing was positive for CSF on beta-2 transferrin assay. The patient denied a history of meningitis, head trauma, otitis media, or prior otologic or neurosurgical procedures. High-resolution temporal bone computed tomography demonstrated significant pneumatization of the temporal bone and marked dilatation and dehiscence of the geniculate fossa (Figure 1). The patient underwent a combined transmastoid–middle cranial fossa exploration with facial nerve monitoring. Intraoperative findings were significant for an enlarged and dehiscent geniculate fossa with marked herniation of the leptomeninges through this fallopian canal defect in the middle fossa floor (Figure 2).

On careful examination, the middle fossa dura mater was found to be intact. The meningocele was opened sharply to relieve the pressure within the dilated facial nerve canal. The area of dehiscence was carefully packed with autologous temporalis fascia and muscle with a DuraGen (Integra Lifesciences Corporation; Plainsboro, New Jersey) overlay. As the dehiscence was packed, there was moderate neurotonic firing from the facial nerve monitor. Care was taken to repair the CSF leak without overpacking and injuring the facial nerve. Postoperatively, no facial weakness developed; however, right-sided high-volume CSF otorrhea persisted. Treatment options were discussed with the patient, including observation with placement of a lumbar drain, attempt at revision packing of the fallopian canal dehiscence via middle cranial fossa with risk of facial nerve injury, subtotal petrosectomy with ear canal closure, and ventriculoperitoneal shunt. Given the high volume of CSF otorrhea immediately after surgery, we estimated the probability that the defect would durably seal with observation or a lumbar drain was extremely low. A ventriculoperitoneal shunt was also offered; however, the patient was counseled that this technique may not resolve the fistula and there would be an unknown persistent risk of meningitis with a shunt. In considering options, collectively the surgical team and patient thought that subtotal petrosectomy with ear canal closure provided the best chance of durable repair with a very low risk of facial nerve injury. The following day, within 20 hours of her original surgery, the patient underwent subtotal petrosectomy with blind-sac closure of the external auditory canal. On postoperative lumbar puncture, an opening pressure of 36 cm H_2O confirmed idiopathic intracranial hypertension (IIH) as a contributing factor to CSF leak. She had no clinical symptoms or signs of elevated intracranial pressure, and therefore a weight loss program was recommended instead of upfront ventriculoperitoneal shunt placement. At 9 months after surgery, the patient has an expected right-sided conductive hearing loss but no evidence of CSF leak, episodes of meningitis, or facial nerve weakness. She has lost 40 kg through dietary modifications after referral to a weight loss clinic.

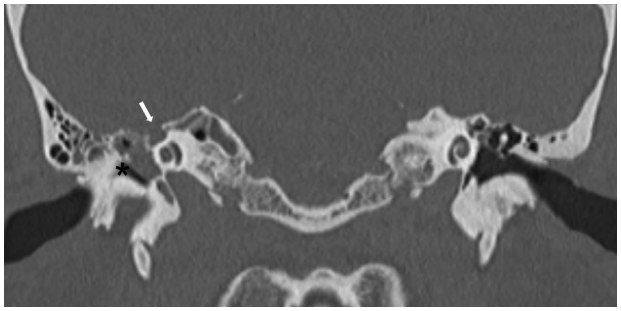

Figure 1. Preoperative high-resolution temporal bone computed tomography coronal reconstruction showing right ear with bony dehiscence over the geniculate fossa (*arrow*) and fluid in the middle ear space (*asterisk*) and normal left ear with intact bone over the geniculate fossa.

Review of Literature

In reviewing the literature, we found 14 prior cases of fallopian canal CSF leak. Table 1 provides a summary of these cases and the present case. The first case was reported in 1967. Age range of patients in the 15 total cases was 2–64 years. The site of fallopian canal dehiscence was the geniculate fossa in 73% (11 of 15) of cases and the tympanic segment in 27% (4 of 15) of cases. Preoperative episodes of meningitis were reported in 53% (8 of 15) of patients. No preoperative facial paralysis was noted. Surgical approaches included transmastoid, middle fossa craniotomy, or a combined transmastoid–middle fossa craniotomy. Materials used for repair varied but in many cases involved packing of the dehiscence with temporalis fascia or muscle. The rate of postoperative facial paralysis was 20% (3 of 15 cases). The rate of persistent CSF leak following initial repair was 27% (4 of 15 cases). Of the 4 cases with postoperative CSF leak, 1 case was managed with revision surgery and repacking of the dehiscence, 1 case was managed with a ventriculoperitoneal shunt (in a patient with IIH), and 2 cases were managed with subtotal petrosectomy with blind-sac closure of the external auditory canal.

DISCUSSION

Fallopian canal meningocele is an exceedingly rare cause of CSF otorrhea. We present a 15th case and review the 14 cases previously reported in the literature to further characterize the clinical presentation, radiographic characteristics, etiology, and management of this rare and enigmatic process.

Clinical Presentation

In reviewing prior cases, there is no clear age or sex predilection. As patients develop CSF leakage from the fallopian canal into the middle ear space, they most commonly present with a unilateral middle ear effusion and conductive hearing loss. This is often

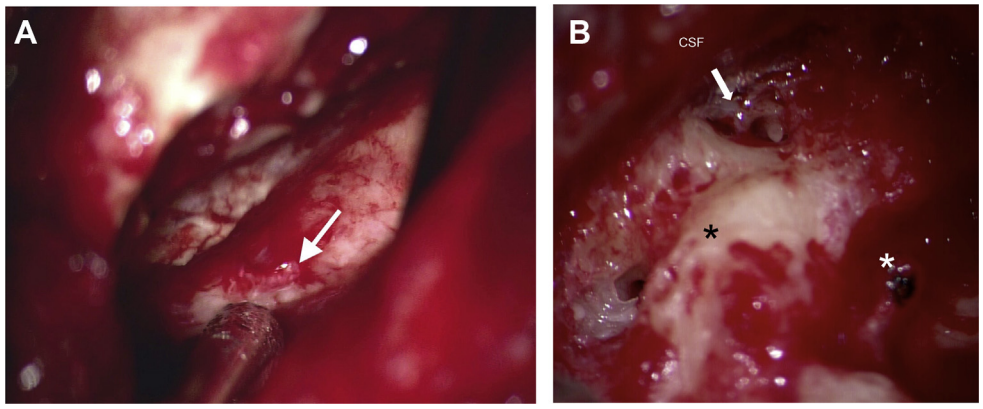

Figure 2. (**A**) Intraoperative photograph from the middle fossa craniotomy approach demonstrating a dehiscent geniculate fossa with herniation of the leptomeninges (*white arrow*). (**B**) Intraoperative photograph from the transmastoid approach illustrating cerebrospinal fluid emanating from a fallopian canal defect (*white arrow*). The horizontal semicircular canal (*black asterisk*) and stapes suprastructure (*white asterisk*) are also shown for orientation. CSF, cerebrospinal fluid.

initially misdiagnosed as serous otitis media. If a myringotomy is performed, persistent otorrhea is typically evident as in the presented index case. If a myringotomy is not performed, patients may present with ipsilateral CSF rhinorrhea via the eustachian tube and/or persistent conductive hearing loss. Preoperative episodes of meningitis were reported in 53% of cases with several patients experiencing recurrent meningitis before receiving an accurate diagnosis and treatment. These patients often have no prior history of head trauma or otologic or neurosurgical procedures. IIH is a risk factor but may not be present in all cases (discussed further below).

Radiographic Characteristics

Classic computed tomography findings of geniculate fossa meningocele include an enlarged geniculate fossa connecting with a widened and shortened labyrinthine segment of the fallopian canal. If magnetic resonance imaging is obtained, a well-circumscribed hyperintense lesion on T2 sequences in the area of fallopian canal dehiscence, consistent with an arachnoid hernia full of CSF, will be seen, as illustrated in **Figure 3**.[12,13] If an active CSF leak is present, the middle ear and mastoid will usually demonstrate partial or complete opacification.

Etiology

The facial nerve is normally sealed from the subarachnoid space by a tight arachnoid band at the meatal foramen. This seal is buttressed by the tight enclosure of the labyrinthine segment of the fallopian canal. It is hypothesized that an abnormally wide labyrinthine segment of the fallopian canal allows the subarachnoid space to extend laterally into the fallopian canal.[5,6,10,14] Gacek[14] described this extension of the subarachnoid space within the fallopian canal in a review of the temporal bone. The constant CSF pressure and pulsations that are allowed to traverse a portion of the fallopian canal by this extension of the subarachnoid space may eventually cause erosion of surrounding bone, resulting in dehiscence and herniation of the leptomeninges. This process may be exacerbated or accelerated by IIH in some patients. Interestingly, not all cases report a geniculate fossa dehiscence in the middle cranial fossa floor as in the present case, but instead report enlargement of the geniculate fossa with thin overlying bone. For example, Foyt and Brackmann[6] described the following: "A large thin, bony bulge was noted over the geniculate ganglion. On removal of a small piece of bone, a copious release of CSF was observed."

In our experience, small asymptomatic facial canal meningoceles are not overly rare; however, active CSF leakage and/or recurrent meningitis is very uncommon, as implied by only 15 total cases reported to date. Although only speculation, it is possible that cases that manifest early in life may result from a more substantial congenital widening of the fallopian canal, whereas cases that manifest later in life, particularly in patients with elevated intracranial pressure, result from a smaller congenital defect and decades of erosive pressure.

Treatment

By understanding the nature by which CSF leaks from a facial nerve meningocele differ from the more common tegmen tympani or mastoideum encephalocele, one can appreciate the unique challenges posed by this enigmatic process. Specifically, because the source of CSF pressure emanates from the posterior fossa, fallopian canal CSF leaks frequently manifest with higher volume flow than typical middle fossa leaks. Furthermore, direct repair of the anatomic defect requires that the surgeon strategically occlude the fallopian canal, without injuring the facial nerve. The prospect of successful repair without facial nerve injury may be particularly challenging in patients with elevated intracranial pressure, such as in the index case presented here.

The elevated risks of refractory CSF leak and postoperative facial paralysis should be discussed with the patient preoperatively. In the 15 reported cases, 27% of patients (4 of 15) experienced

Table 1. Summary of All Previously Published Cases of Fallopian Canal Meningocele with Cerebrospinal Fluid Leak

Reference	Age (Years)/Sex	Meningitis	Site of Fallopian Canal Dehiscence	Surgical Approach	Repair of Dehiscence	Postop Facial Paralysis	Postop CSF Leak	Secondary Repairs
Harrington and Brik, 1967[1]	9/F	Yes	Geniculate ganglion	TM	Bone chips, absorbable gelatin sponge	No	Yes	Revision transmastoid approach, repacking with gelatin sponge, middle ear obliteration with temporalis muscle
Gacek and Leipzig, 1979[2]	2/M	Yes	Tympanic segment	TM	Temporalis fascia	No	No	—
Barcz et al., 1985[3]	14/M	Yes	Tympanic segment	TM	Temporalis fascia	Yes	No	—
Legent et al., 1989[4]	7/M	Yes	Geniculate ganglion	TM	Temporalis fascia, bone pate, fibrin glue	No	No	—
Petrus and Lo, 1999[5]	34/M	Yes	Geniculate ganglion	MFC	Temporalis muscle	No	No	—
	5/M	No	Geniculate ganglion	MFC	Temporalis muscle	No	No	—
Foyt and Brackmann, 2000[6]	34/M	Yes	Geniculate ganglion	TM, MFC	Temporalis muscle	Yes	No	—
	5/M	No	Geniculate ganglion	TM, MFC	Temporalis muscle, Surgicel*	No	No	—
Piane et al., 2001[7]	64/F	No	Tympanic segment	TM	Temporalis muscle, Tisseel†, abdominal fat in mastoid	No	No	—
Isaacson et al., 2002[8]	37/M	Yes	Tympanic segment	TM	Temporalis muscle, bone pate, fibrin glue	No	Yes	Revision transmastoid approach, repacking of fallopian canal; CSF leak persisted, eventually treated with subtotal petrosectomy with blind-sac closure of EAC
Mong et al., 2009[9]	25/F	No	Geniculate ganglion	MFC	Temporalis muscle, Tisseel†, temporalis fascia, bone graft	No	Yes	Ventriculoperitoneal shunt
	7/M	No	Geniculate ganglion	MFC	Temporalis muscle, temporalis fascia, bone graft	Yes	Not from geniculate ganglion (leaked from other anterior skull base dehiscence)	
Dhanasekar et al., 2010[10]	9/NR	Yes	Geniculate ganglion	TM	Not repaired primarily; subtotal petrosectomy with blind-sac closure of EAC in conjunction with bilateral cochlear implants for postmeningitic SNHL	No	No	
Teufert and Slattery, 2013[11]	45/F	No	Geniculate ganglion	TM, MFC	Temporalis muscle	No	No	
Dey et al., 2018 (present study)	53/F	No	Geniculate ganglion	TM, MFC	Temporalis fascia and muscle with DuraGen overlay	No	Yes	Subtotal petrosectomy with blind-sac closure of EAC

Postop, postoperative; CSF, cerebrospinal fluid; F, female; TM, transmastoid; M, male; MFC, middle fossa craniotomy; EAC, external auditory canal; NR, not reported; SNHL, sensorineural hearing loss.

*Surgicel (Ethicon Inc., Somerville, New Jersey, USA).

†Tisseel (Baxter international Inc., Deerfield, Illinois, USA).

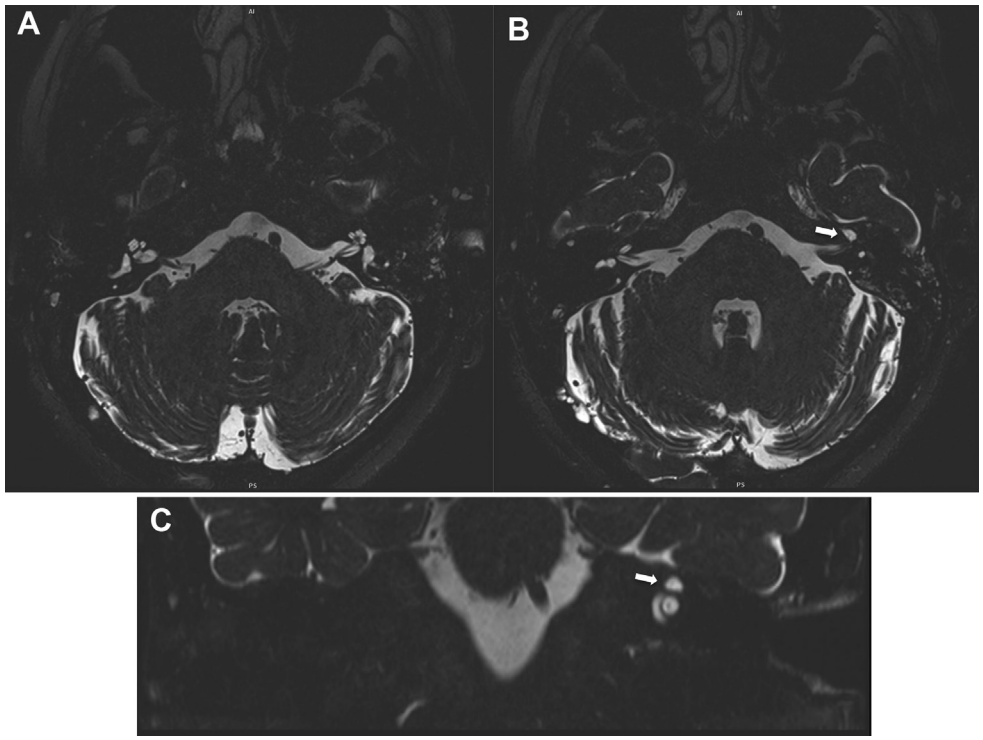

Figure 3. Separate illustrative case of a patient previously evaluated by the authors with an incidental fallopian canal meningocele involving the geniculate fossa and middle ear. Thin-slice heavily weighted T2 magnetic resonance imaging in the axial (**A** and **B**) and coronal (**C**) planes demonstrating a 4 × 3 × 2 mm meningocele positioned just lateral to the geniculate fossa and superior to the cochlea (*white arrow* in **B** and **C**).

persistent postoperative CSF leak, and 20% of patients (3 of 15) experienced postoperative facial nerve paralysis. The 3 cases with postoperative facial paralysis were reviewed. Barcz et al.[3] did not report a postoperative facial paralysis grade, but they reported the patient's facial function was slowly improving at follow-up. The patient reported by Foyt and Brackmann[6] experienced House-Brackmann grade II/VI facial paralysis postoperatively, which recovered to grade I/VI after several weeks. The case reported by Mong et al.[9] had House-Brackmann grade VI/VI facial paralysis postoperatively that recovered to grade III/VI.

Most cases (73%) were from a dehiscence located in the geniculate fossa. Most of these cases were approached via a middle fossa craniotomy or combined transmastoid–middle fossa craniotomy. For the cases with a tympanic segment dehiscence, a transmastoid approach was sufficient. As described in **Table 1**, various materials have been used to pack around the dehiscent fallopian canal. Temporalis fascia and muscle were most commonly used.

Several of the patients who experienced refractory CSF leak were reported to have IIH. Intracranial hypertension further challenges a successful repair, and the need for revision surgery should be discussed with patients suspected to have this comorbid condition. In cases of refractory CSF leak, at least 4 options are available: observation and placement of a lumbar drain, revision middle fossa craniotomy and repacking of the dehiscence, ventriculoperitoneal shunt, and subtotal petrosectomy with blind-sac closure of the external auditory canal.

In cases of highovolume refractory CSF leak, we believe that observation with lumbar drain placement is unlikely to facilitate a durable and permanent seal of the defect, particularly in patients with suspected IIH. Choice among the remaining 3 options is largely based on discussion with the patient and the risks the patient is willing to accept. Often, patients are not willing to accept significant risk to the facial nerve or the possibility of persistent CSF otorrhea. In these cases, subtotal petrosectomy with blind-sac closure of the external auditory canal warrants strong consideration. With this procedure, the surgeon and patient are effectively trading a maximal conductive hearing loss, which can be rehabilitated with a bone-anchored hearing aid, for the benefit of definitive repair and minimal risk to the facial nerve. This technique was also used by Isaacson et al.[8] to manage a fallopian canal CSF leak that recurred multiple times.

CONCLUSIONS

Fallopian canal meningocele is an exceedingly rare cause of CSF otorrhea, with only 15 cases now reported in the literature. These CSF leaks are difficult to treat with increased risk of postoperative facial paralysis and refractory CSF leak. For refractory CSF otorrhea, treatment options include observation with lumbar drain

placement, revision packing of the fallopian canal dehiscence, subtotal petrosectomy with closure of the external auditory canal, and ventriculoperitoneal shunt placement. In cases of refractory CSF leak with concomitant IIH, subtotal petrosectomy with closure of the external auditory canal provides durable repair with minimal risk of facial nerve injury.

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