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Management of spontaneous temporal bone cerebrospinal fluid leak: A 30-year experience☆

Esther Cheng[a,*], David Grande[b], John Leonetti[a]

[a] Department of Otolaryngology – Head & Neck Surgery, Loyola University Medical Center, Maywood, IL, United States of America
[b] Loyola University Chicago Stritch School of Medicine, Maywood, IL, United States of America

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ABSTRACT

Purpose: The goal of this study was to describe the incidence of and our long-term results in the management of spontaneous temporal bone CSF leak and to determine if there is any association with an increase in body mass index (BMI).

Methods: This was an Institutional Review Board (IRB) approved retrospective review of 58 patients who underwent repair of spontaneous temporal bone CSF leaks by a single surgeon at a tertiary care academic medical center between 1988 and 2017. Patients with CSF leaks due to trauma, iatrogenic injury, or chronic infection were excluded. Surgical approaches included middle cranial fossa, transmastoid, or a combination of both. Cases were divided into three decades. Patient demographics, clinical features, audiologic results, imaging studies, operative findings, complications, and long-term results were reviewed with a specific emphasis on the potential correlation with BMI.

Results: The average BMI was 32. There was a statistically significant increase in BMI between the two most recent decades ($p = 0.044$). The middle cranial fossa was the most commonly utilized approach. Multiple tegmen defects were often noted on imaging and intraoperatively.

Conclusion: There is an increase in the incidence of spontaneous CSF leaks possibly associated with an increase in BMI in the United States over the last 30 years.

1. Introduction

Temporal bone cerebrospinal fluid (CSF) leaks most commonly occur secondarily as a result of trauma, chronic otitis media, or a previous surgery; however, spontaneous CSF leaks have become increasingly more prevalent in recent years [1]. While traumatic temporal bone CSF leaks tend to heal with conservative management, idiopathic CSF leaks typically require surgical intervention. The three primary surgical approaches are transmastoid, middle cranial fossa, and a combination of the transmastoid and middle cranial fossa approaches.

Kari et al. noted that BMI in patients with spontaneous CSF leaks was significantly higher than that of patients with CSF leaks due to traumatic, infectious or iatrogenic etiologies [2]. There are a number of theories regarding the etiology of spontaneous temporal bone CSF leaks including congenital defects and arachnoid granulation [3,4]. However, recent studies have implicated a role of obesity [1,2,4]. There has been a perceived increase in the number of CSF leaks in our patients over the past three decades. The main purpose of the study is to describe our long-term results following the surgical repair of spontaneous temporal bone CSF leaks over three decades in a single institution and provide further elucidate the role of obesity in development of CSF leaks.

2. Patients and methods

An Institutional Review Board (IRB) approved retrospective review was conducted on patients with spontaneous temporal bone CSF leaks who underwent surgical repair by a single surgeon at Loyola University Medical Center from 1988 to 2017. Patients with CSF leaks resulting from trauma, surgery, or infection were excluded. A total of 58 patients met inclusion criteria. Patient demographics were recorded including gender, age, body mass index (BMI), and medical comorbidities. Physical exam, audiologic, and imaging findings were noted. Surgical repair was performed using a transmastoid, middle cranial fossa (MCF), or combined approach. Transmastoid repair was accomplished with

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* Corresponding author at: Loyola University Health System, Department of Otolaryngology, Maguire Center Room 1870, 2160 South First Ave, Maywood, IL 60153, United States of America.
E-mail address: Esther.Cheng@luhs.org (E. Cheng).

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temporalis fascia and auricular cartilage for the dural and tegmen defects respectively. Middle cranial fossa repairs were performed also with a temporalis fascia graft in addition to a split thickness inner table bone graft and fibrin glue for the tegmen defect. Combined transmastoid and MCF repairs were accomplished in the same manner as the MCF approach alone.

We initially sought to evaluate patients across three decades; however, most of the records from the first decade were incomplete, and many of the cases were iatrogenic, infectious, or traumatic in nature. As there was only one patient that met criteria in the first decade, that data was not included in the BMI trend analysis.

Statistical analysis was performed on BMI data using simple *t*-test. Where applicable, the *p*-value was significant if < 0.05. Presenting symptoms, physical examination findings, surgical approach, and complications were presented as counts or percentages.

3. Results

A total of 58 patients were reviewed. The average age was 58 with a range of 14 to 84. There were equal numbers of male and female patients. The number of spontaneous CSF leaks per decade was 1, 21, and 36 (Table 1). The overall average BMI for this series was 32 with a range of 20.7 to 46.7. The average BMI for the second and third decades was 29.7 and 33.3 ($p = 0.0044$) (Table 1) (Fig. 1). Average BMI for the first decade was not included as there was only one patient that fit the inclusion criteria. The most common medical comorbidity was hypertension ($n = 35$) followed by diabetes mellitus ($n = 13$).

The most common presenting symptoms were otorrhea after tympanostomy tube placement (67.2%) and ear fullness (50%) (Table 2). Presence of a ventilation tube with clear otorrhea was the most frequent physical exam finding (56.9%) with middle ear effusion being the second most common (44.8%).

All patients underwent evaluation with a preoperative audiogram and computed tomography (CT) scan of the temporal bone. Mixed hearing loss was the most frequent audiologic finding on the affected side. CT scans demonstrated one or more tegmen defects (Fig. 2). The finding of an encephalocele was less common (34.5%). In the rare instance that CT findings were unclear, an MRI was obtained (12%). Beta-2-transferrin was never ordered in this series.

The most frequently utilized surgical approach was the middle fossa approach (79.3%) (Table 3). When the location of the leak was noted in the operative report, the most common location was the tegmen tympani, although 30.3% of cases had defects in both the tegmen tympani and mastoideum. The majority of the leaks originated from the right ear (56.9%). Concurrent encephalocele was noted in 91.3% of patients (Fig. 3). One large tegmen defect was seen in 62% of cases. The remainder of cases either had multiple small tegmen defects or a combination of one large and multiple small defects. There were no negative explorations in this series.

If complications did occur (13.8%), they were most frequently delayed in nature (Table 4). Only three patients experienced early complications in the immediate postoperative period. The most common complication was persistent CSF leak (5.2%). Two patients were initially managed with a lumbar drain. One leak persisted despite the lumbar drain and resolved after a revision transmastoid approach with ear canal oversew and Eustachian tube obliteration. One patient was taken immediately to the operating room for repair via a transcochlear approach with a temporalis muscle rotational flap and abdominal fat graft. This leak was considered more brisk and unlikely to resolve with a lumbar drain. All three cases of persistent CSF leak occurred in cases where the middle fossa approach was used, though this was overall the most commonly utilized approach. Persistent leaks were due to very small tegmen defects that were located anteriorly. There were no CSF leaks that recurred in a delayed fashion.

Table 1
Average BMI and number of CSF leaks per decade studied.

Decade	1988–1997	1998–2007	2008–2017	p = 0.044
Number of CSF leaks	1	21	36	
Average BMI	–	29.7	33.3	

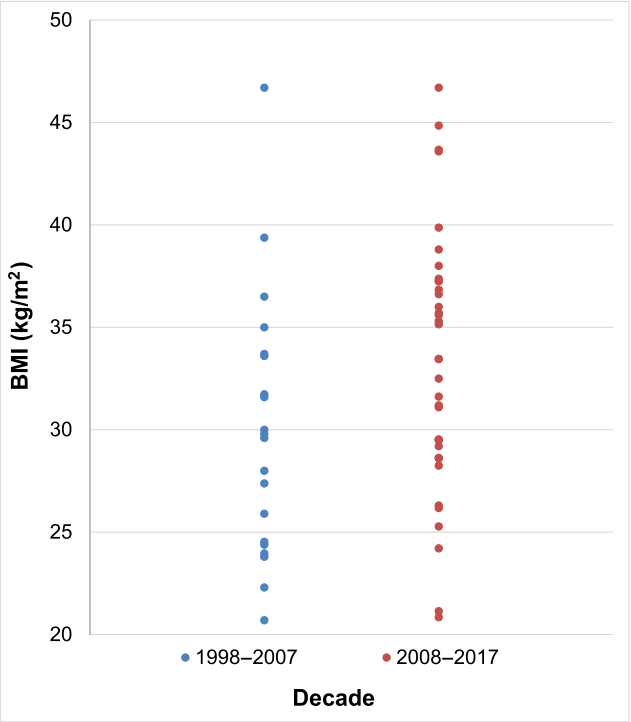

Fig. 1. This is a scatter plot demonstrating the BMI distribution across the decades.

Table 2
Presenting symptoms.

Symptom	Incidence
Hearing loss	37.9%
Ear fullness/pressure	50%
Pulsatile tinnitus	24.1%
Otorrhea	67.2%
Headache	8.6%

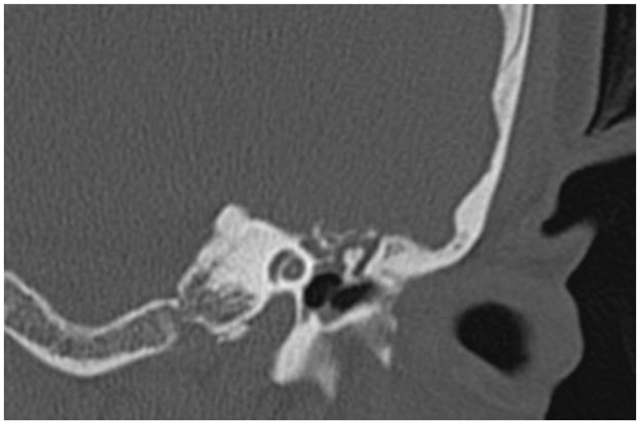

Fig. 2. This is a coronal CT demonstrating multiple tegmen defects in the left temporal bone.

Table 3
Surgical approach used for repair.

Approach	# of cases	# of persistent leaks	# of recurrent leaks
Middle cranial fossa (MCF)	47	3	0
Transmastoid (TM)	4	0	0
Combination MCF/TM	7	0	0

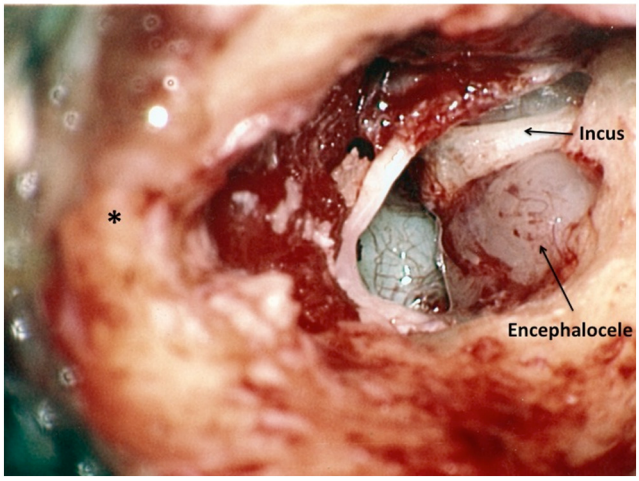

Fig. 3. This intraoperative photograph during a combined transmastoid and middle cranial fossa approach demonstrates a left ear with visualized encephalocele in the middle ear abutting the incus. The asterix indicates inferior.

Table 4
Complications from surgery.

Complication	#
Hematoma	1
Wound infection	2
Wound dehiscence	1
Persistent CSF leak	3
Delayed facial paralysis	1

4. Discussion

Spontaneous temporal bone CSF leaks are less common than leaks with a discernable etiology, though they are increasing in frequency. The etiology of spontaneous temporal bone leaks has been attributed to tegmen atrophy as a result of erosion from CSF pulsations, arachnoid granulations, and congenital bone reabsorption [1].

Spontaneous temporal bone CSF leaks are thought to be more common in obese patients due to persistent increased intra-abdominal pressure, which decreases venous return and increases intracranial pressure [5]. Average BMI in numerous studies of patients with spontaneous temporal bone CSF leaks ranged from 33.2–40.7 [1,4–7]. This is well above the national average of 26.5 and within the obese BMI category. The average BMI in our study was 32 with a range of 20.7 to 46.7. Average BMI for the second and third decades were 29.7 and 33.3. The observed increase in average BMI reached statistical significance. Other studies have demonstrated a higher average BMI in patients with spontaneous temporal bone CSF leaks when compared to patients with CSF leaks of other etiologies [7,8].

While we did not have BMI data for the first decade studied (1988–1997), we found that only one out of eight cases was a spontaneous CSF leak. The remainder of the cases had an identifiable cause. This provides further argument for the increase in incidence of spontaneous CSF leaks in recent years, whether it is an increase in true incidence or an increase in awareness.

Some studies have noted a higher female to male ratio in the development of spontaneous temporal bone CSF leaks, while others did not observe this trend [1,4,6,9]. We did not find gender predominance in the diagnosis of spontaneous temporal bone CSF leak. Right-sided predominance of CSF leaks was noted in several studies [9]. This finding was replicated in our study.

Common presenting symptoms and signs include aural fullness, hearing loss, pulsatile tinnitus, and recurrent middle ear effusion [4]. CSF leak is often suspected when there is persistent clear otorrhea after tympanostomy tube insertion [4,6,10]. This was the most common finding in our series. History and physical examination alone are often sufficient for diagnosis; however, a high-resolution computed tomography (CT) scan of the temporal bone with coronal cuts is usually recommended for locating tegmen defects.

The use of beta-2-transferrin testing remains controversial as CSF leaks can often be diagnosed with clinical history and examination alone often in conjunction with imaging. Gonen et al. found only a 55% rate of beta-2-transferrin positivity, while Kim et al. report at 75% rate of positivity. The high false negative rate has been attributed to slow and intermittent CSF leaks [4]. Many authors including the senior author for this study did not routinely obtain beta-2-transferrin if there was high enough suspicion based on history and physical and imaging findings. There were no negative explorations in this series, suggesting that obtaining beta-2-transferrin may be unnecessary.

Anatomic studies have demonstrated a rate of single to < 5 tegmen defects between 15 and 34% and a rate of multiple tegmen defects of < 1%, though that number is rising [11,12]. Numerous studies reported multiple tegmen defects intra-operatively at rates ranging from 25 to 56% [1,7,10,13]. This was similar to the findings in our study, which demonstrated a 58.6% rate of multiple tegmen defects. Concurrent encephalocele was noted in several but not all patients with CSF leaks (83.3%).

Multiple surgical approaches have been used for repair. The middle cranial fossa approach was most commonly used in our study, and while this is the only approach associated with persistent leaks in this series, it was by far the most commonly used approach. Therefore, recurrences would not be unexpected. A transmastoid approach has been recommended for defects < 1 cm that are limited to the tegmen mastoideum, whereas a MCF approach is recommended for larger defects involving the tegmen tympani. A combined transmastoid and MCF approach has been used in multiple studies with very high success rates attributed to better exposure of medial, multiple, and large defects [1,4,6]. Recurrent leaks tended to be more common in cases where a transmastoid approach alone was used [1]. Recurrences were approached either via a middle craniotomy or a combination of the transmastoid and MCF approaches [5].

Persistent CSF leak was the most common complication after surgery followed by wound infection. Delayed facial paralysis occurred in one patient who underwent a MCF approach. This was presumably caused by stretching of the greater superficial petrosal nerve during elevation of the temporal lobe dura. The facial paralysis completely resolved one month after surgery. The patient did receive a three-week prednisone taper.

Our study demonstrates an overall increase in the incidence of spontaneous temporal bone CSF leaks. This trend mirrors that of average BMI per decade studied. Gonen et al. also observed a similar trend where 76% of the 245 reported spontaneous temporal bone CSF

leaks were noted in more recent years (2000 − 2013) compared to earlier years (1986–1999). The increase in number of spontaneous temporal bone CSF leaks has been attributed to an increase in awareness as well as a true increase in incidence. The incidence of obesity has increased as well [1]. It is possible that there has also been an increase in surgical referrals to this tertiary care institution for spontaneous temporal bone CSF leaks. As the number of patients in this series increased, the average BMI increased as well. Additional studies are necessary to investigate true association; however, there is a trend of increased incidence of spontaneous temporal bone CSF leaks that matches the increase in BMI in the United States.

5. Conclusions

The incidence of spontaneous temporal bone CSF leaks has increased in recent years. In this series, the average BMI increased per decade studied. The increased incidence of spontaneous temporal bone CSF leaks may be attributed to an overall increase in average BMI in the United States.

References

[1] Gonen L, Handzel O, Shimony N, Fliss DM, Margalit N. Surgical management of spontaneous cerebrospinal fluid leakage through temporal bone defects—case series and review of the literature. Neurosurg Rev 2016;39:141–50.

[2] Kari E, Mattox DE. Transtemporal management of temporal bone encephalocele and CSF leaks: review of 56 consecutive patients. Acta Otolaryngol 2011;131:391–4.

[3] Markou K, Goudakos J, Franco-Vidal V, Vergnolles V, Vignes JR, Darrouzet V. Spontaneous osteodural defects of the temporal bone: diagnosis and management of 12 cases. Am J Otolaryngol 2011;32:135–40.

[4] Kim L, Wisely CE, Dodson EE. Transmastoid approach to spontaneous temporal bone cerebrospinal fluid leaks: hearing improvement and success of repair. Otolaryngol Head Neck Surg 2014 Mar;150(3):472–8.

[5] Kutz Jr. JW, Husain IA, Isaacson B, Roland PS. Management of spontaneous cerebrospinal fluid otorrhea. Laryngoscope 2008;118:2195–9.

[6] Kenning TJ, Willcox TO, Artz GJ, Schiffmacher P, Farrell CJ, Evans JJ. Surgical management of temporal meningoencephaloceles, cerebrospinal fluid leaks, and intracranial hypertension: treatment paradigm and outcomes. Neurosurg Focus 2012;32(6):1–8.

[7] Son HJ, Karkas A, Buchanan P, Giurintano JP, Theodosopoulos P, Pensak ML, et al. Spontaneous cerebrospinal fluid effusion of the temporal bone: repair, audiological outcomes, and obesity. Laryngoscope 2014;124:1204–8.

[8] Stucken EX, Selesnick SH, Brown KD. The role of obesity in spontaneous temporal bone encephaloceles and CSF leak. Otol Neurotol 2012;33:1412–7.

[9] Nahas Z, Tatlipinar A, Limb CJ, Francis HW. Spontaneous meningoencephalocele of the temporal bone. Arch Otolaryngol Head Neck Surg 2008;134(5):509–18.

[10] Semaan MT, Gilpin DA, Hsu DP, Wasman JK, Megerian CA. Transmastoid extradural-intracranial approach for repair of transtemporal meningoencephalocele: a review of 31 consecutive cases. Laryngoscope 2011;121:1765–72.

[11] Kapur TR, Bangash W. Tegmental and petromastoid defects in the temporal bone. J Laryngol Otol 1986;100(10):1129–32.

[12] Lang DV. Macroscopic bony deficiency of the tegmen tympani in adult temporal bones. J Laryngol Otol 1983;97(8):685–8.

[13] Gubbels SP, Selden NR, Delashaw Jr. JB, McMenomey SO. Spontaneous middle fossa encephalocele and cerebrospinal fluid leakage: diagnosis and management. Otol Neurotol 2007;28:1131–9.