

# Endonasal Endoscopic Repair of Spontaneous Cerebrospinal Fluid Leaks

Andrey S. Lopatin, MD; Dmitry N. Kapitanov, MD; Alexander A. Potapov, MD

**Objective:** To analyze possible etiological factors of spontaneous cerebrospinal fluid (CSF) rhinorrhea and to assess the outcomes of endonasal endoscopic repair.

**Design:** Retrospective study.

**Setting:** Academic neurosurgical hospital.

**Patients:** Twenty-one consecutive patients who presented with spontaneous CSF leak and underwent endonasal endoscopic surgery from January 1999 through December 2001.

**Intervention:** Preoperative examination included computed tomographic scans; nasal endoscopy; measurement of glucose concentration in the nasal discharge; and, in some cases, cisternographic evaluations via computed tomography and/or magnetic resonance imaging. Telescopes, conventional endoscopic sinus surgery instruments, and a microdebrider were used for all patients who underwent endonasal surgery. A combination of plastic materials, ie, abdominal fat, fascia lata, rotated middle turbinate flaps, and fibrin glue, were used for fistula repair.

**Results:** At the time of surgery, CSF fistulas were found

in the cribriform plate (6 patients), in the fovea ethmoidalis (6 patients), and in the sphenoid sinus (9 patients). In 5 of the 6 patients who had an extremely pneumatized sphenoid sinus, the source of the leak was located in the lateral extension of the sinus. A meningocele protruding through the bone defect was the source of the leak in 10 patients. Postoperative follow-up lasted from 9 to 42 months, and 20 patients were considered cured. There was only 1 recurrence, in a patient whose CSF rhinorrhea originated in the deep lateral recess of an overpneumatized sphenoid sinus. Thus, the overall success rate was 95.2%. There were no postoperative complications.

**Conclusions:** Possible etiological factors of this disease include obesity, congenital malformations of the skull base, an overpneumatized sphenoid sinus (particularly in its lateral extensions), and the empty sella syndrome. Endoscopic endonasal repair of spontaneous CSF rhinorrhea appears to be a safe and successful procedure. However, techniques for endoscopic closure of CSF fistulas in the lateral part of the sphenoid sinus need further perfecting.

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*From the Ear, Nose, and Throat Department, Central Hospital, Presidential Medical Center (Dr Lopatin), and the Burdenko Institute of Neurosurgery, Russian Academy of Medical Science (Drs Kapitanov and Potapov), Moscow, Russia. The authors have no relevant financial interest in this article.*

**P**ERSISTENT CEREBROSPINAL fluid (CSF) leaks are usually considered primary (spontaneous) or secondary. Secondary CSF leaks are rather common and accompany 80% of severe head traumas. A specific form of secondary traumatic CSF leak is iatrogenic liquorrhea, which occurs not infrequently during skull base and endonasal sinus surgery. Secondary CSF rhinorrhea can also be caused by skull base and intracranial tumors such as angiofibromas and osteomas, which destroy skull base bones. In contrast, primary CSF rhinorrhea has been usually recognized as a rare condition presenting diagnostic and therapeutic challenges.

Developments in endonasal endoscopic surgery have made endonasal pro-

cedures less invasive, and CSF rhinorrhea is now one of its well-established indications.<sup>1,2</sup> In most cases, endoscopic nasal surgery for CSF leak repair has almost completely replaced more traumatic transcranial and extracranial procedures. However, methods for the correct location and techniques for the surgical closure of spontaneous CSF fistulas need further perfecting.

## METHODS

### PATIENTS

Of the 69 patients who were admitted from January 1999 through December 2001 to the Institute of Neurosurgery and diagnosed with CSF rhinorrhea, 30 had posttraumatic and 14 had postoperative leaks. A diagnosis of spon-

taneous CSF rhinorrhea was confirmed in the remaining 25 patients. Of these, 4 were operated on with the transcranial approach and 21 underwent endonasal endoscopic surgery. The 15 women and 6 men who formed the latter group (our study group) were between 15 and 64 years old and only 5 of them (4 women and 1 man) were obese. Of the 21 patients, 5 reported temporary CSF leak from one nasal cavity whereas liquorrhea was permanent in the other 16 patients; and 19 patients had both anterior and postnasal CSF discharge whereas 2 patients who did not report release of CSF from the nostrils experienced permanent leakage from the nasal cavity onto the posterior pharyngeal wall, which caused irritation and cough.

More than one half of the patients (11) in our series experienced at least 1 episode of meningitis, and 4 patients reported recurrent episodes. There were 2 cases of pneumatocephalus. The duration of the condition, ie, the period between first presentation of the CSF leak and admittance to the clinic, was less than 2 months in 2 cases, between 2 and 6 months in 7 cases, and between 6 and 12 months in 4 cases. Eight patients experienced liquorrhea for more than 1 year. Surgical fistula closure had been previously attempted in 6 cases involving 5 patients. In 4 of the patients, it was with lumboperitoneal shunting whereas the fifth patient had 2 operations with the transcranial approach. One patient previously had 2 endonasal and 1 transcranial operations in combination with lumboperitoneal shunting. None of these attempts achieved steady positive results.

Preoperative examination included high-resolution computed tomographic (CT) scans of the paranasal sinuses and skull base, nasal endoscopy, and evaluation of glucose concentration in the nasal discharge. When initial CT scans did not identify a bone defect in the skull base, patients underwent cisternographic evaluation via CT and/or magnetic resonance imaging. Preoperative nasal endoscopic examination with the patient in the supine position located a CSF fistula in 1 patient. In the other patients, the examination revealed which nasal cavity had a wetter and shinier nasal mucosa and permitted collection of transparent fluid in the nasopharynx, but did not allow precise visualization of the source of liquorrhea. Glucose concentration in the nasal discharge was higher than 30 mg/dL (1.7 mmol/L) in all patients.

Computed tomography and cisternographic evaluation via CT or magnetic resonance imaging were able to detect the precise site of the fistula in 17 cases. In the 4 remaining cases, the bone defect itself was not visualized; however, local mucosal edema caused by irritation, collection of CSF in specific sinuses, or ethmoidal cells suggested the fistula's location. A common finding in this series was extremely pneumatized sphenoid sinuses that extended to the apex of the temporal pyramid (**Figure 1**). In 5 cases, a bone defect and/or a small meningocele appearing to be the source of the CSF leak were found in the lateral wall of the overpneumatized sphenoid sinus.

#### SURGICAL TECHNIQUE

Fistula repair was performed with the endonasal endoscopic approach under general anesthesia. In all, 22 operations (21 primary attempts and 1 revision) were carried out over the 3-year period. Lumbar puncture was always performed in the operating room immediately before the beginning of surgery. Cerebrospinal pressure was measured and a lumbar drain was placed—with care taken to avoid evacuation of a significant amount of CSF. During surgery, the drain was kept closed until the final stage of the fistula repair. This maintained the release of CSF into the nasal cavity and thus facilitated the fistula's location.

The surgical technique was dependent on the size and position of the fistula. The operation began with a careful endo-



**Figure 1.** Axial (A) and coronal (B) computed tomographic scans from a patient with cerebrospinal rhinorrhea originating from a lateral extension of the left sphenoid sinus. Note the fluid level (A) and a small meningocele in the lateral part of the sinus (B).

scopic examination of the nasal cavity. Once the patient was in the supine position, examination often revealed fluid collection in the nasopharynx. In cases of rapid CSF leaks, preoperative decongestion of the nasal mucosa and tractions on the middle turbinate identified a CSF pathway, either from the natural ostium of the sphenoid sinus, or from the natural ostium of the posterior ethmoid sinus, or from the posterior part of the middle meatus on the upper wall of the choana. In cases where CT data were not conclusive, immediate preoperative endoscopy permitted the approximate location of the bone defect and the selection of a tentative surgical technique.

In no case did the operation begin with resection of the middle turbinate. Fracture and maximal lateral rotation of the turbinate were enough for good visualization of a bone defect in the medial portion of the cribriform plate. For fistulas located in the lateral part of the cribriform plate and in the fovea ethmoidalis we rotated the middle turbinate and cut a wide opening in the ethmoid sinus using the straight 3.5-mm blade of a microdebrider (Hummer 2; Stryker Endoscopy, Santa Clara, Calif). The steps for fistulas in the sphenoid sinus were lateralization of the middle turbinate; identification of the sphenoidal ostium; and its enlargement inferiorly and laterally by cutting downward with the 3-mm, 90° downward Kerrison punch (Leibinger GmbH, Freiburg, Germany). A CSF fistula could not be easily approached in the lateral wall of the overpneuma-

**Location of CSF Fistulas and Results of Surgical Repair**

Location of CSF Fistulas	Total No. (Leak Associated With Meningocele)	Recurrence and Revision Surgery
Cribriform plate	6 (5)	None
Roof of anterior ethmoid sinus	2 (0)*	None
Roof of posterior ethmoid sinus	2 (1)	None
Roof of ethmoid sinus (without precise intraoperative location)	2 (0)	None
Lateral recess of sphenoid sinus	5 (4)	1
Posterior or superior wall of sphenoid sinus	4 (0)	None
<b>Total</b>	<b>21 (10)</b>	<b>1</b>

\*Precise intraoperative location of the fistula was not possible at the time of surgery.

tized sphenoid sinus; but even in that case, middle turbinate resection was not performed because this would not have provided any benefit. If needed, the turbinate was returned to its initial medial position for better visualization and easier access to the lateral part of the sinus.

It is imperative for successful fistula repair to remove the mucosa from the edges of the bone defect. In the case of a meningocele, the latter was removed and the bone edges of the fistula were carefully skeletonized. Selection of a plastic material for fistula repair was dependent on the size and position of the fistula. If the bone defect was precisely visualized, several layers of abdominal fat were placed inside the defect, which was then closed with a piece of autogenous fascia lata in an on lay fashion. Middle turbinate flaps were rotated for the closure of bone defects in the cribriform plate and fovea ethmoidalis. If direct visualization of the fistula with a 0° endoscope was not possible, the walls of the diseased sinus were deepithelialized as much as possible and the sinus was obliterated with autogenous fat. Obliteration of the cavity of the ethmoidal cells and medial displacement of the middle turbinate were performed in a way that did not obstruct the frontal sinus ostium. Septal cartilage was used exclusively for closure of large bone defects in the roofs of the ethmoidal sinuses. The plastic materials used are the following:

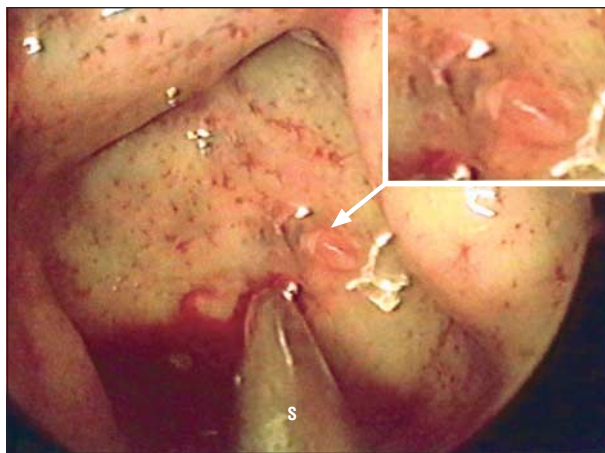
Autogenous Materials Used in Fistula Repair	No.
Abdominal fat	17
Facia lata	15
Rotated middle turbinate flap	11
Nasal septum mucosal flap	1
Septal cartilage	3
Muscle	1

In most patients fistula repair was achieved with a combination of plastic materials, and 0.5 to 1.0 mL of Tissucol fibrin glue (Immuno, Vienna, Austria) was used for fixation of the flaps and plastic materials in all but 1 patients. Two or 3 pieces of Merocel packs (XOMED, Jacksonville, Fla) were placed in the nasal cavity at the end of the procedure.

Patients were allowed to stand on the day after surgery but they were asked to avoid blowing their noses and coughing. The lumbar drain was kept for 5 to 8 days and CSF was evacuated by the drain at the rate of 5 mL per hour. Packs were removed from the common nasal meatus on the third day and from the upper and middle meatus on the fifth or sixth day.

**RESULTS**

The locations of CSF fistulas in this series of patients are listed in the **Table**. In 6 patients the bone defect was found



**Figure 2.** Intraoperative view with a 0° scope. One of the 2 leaking fistulas in the posterior sphenoid sinus wall is under the suction tip (S) and the other is indicated by the arrow.

in the cribriform plate, and the fovea ethmoidalis was the source of CSF rhinorrhea also in 6 patients. Among the latter, the defect was found in the anterior part of the ethmoidal roof in 2, and in the posterior part of the ethmoidal roof in another 2; in the remaining 2 patients, precise intraoperative location of the fistula was not possible because of the absence of active liquorrhea at the time of surgery. In the 9 other patients, CSF rhinorrhea originated from the sphenoid sinus; among these, a fistula was found on the posterior or superior wall of the sinus in 4 patients, and in the overpneumatized lateral part of the sinus in the remaining 5. An extremely pneumatized sphenoid sinus was a common finding. This type of pneumatization was present in 6 patients, and in 5 of these the source of the leak was found in the lateral sinus wall. A meningocele protruding through the bone defect was detected in 10 patients: in 4, it was located in the lateral wall of the sphenoid sinus (always in the lateral recess); and in 6, it was located on the upper wall of the ethmoid sinus.

In 8 patients, the size of the skull base bone defect was not greater than 3 mm; in 4 patients it was between 4 and 5 mm; and in another 4 patients it was larger, between 7 and 12 mm. One patient had 2 small fistulas in the very thin posterior wall of a large sphenoid sinus extending beneath the sella turcica (**Figure 2**). The absence of an active CSF leak at the time of surgery and/or the bone defect's hidden site prevented direct visualization of the fistula and estimation of its size in 4 patients.

Follow-up varied from 9 to 42 months. Immediate results were good in all patients, but later in the follow-up 1 female patient developed recurrent CSF rhinorrhea. In her case the source of the leak, which was situated in a deep lateral recess of the sphenoid sinus, was not appropriately reached and managed during the surgical intervention. As a result, obliteration of the sphenoid sinus was performed instead of dura defect repair. Revision surgery was attempted 9 months later in the same fashion, but obliteration failed again, probably because complete mucosa removal was not possible in the extremely pneumatized lateral recess of the sinus. However, the patient reported improvement (a reduction in

the amount of CSF released and an absence of meningitis). Until now, she has refused further surgical treatment.

#### COMMENT

Head trauma is the most common cause of CSF leak and most trauma-related CSF leaks resolve without surgical intervention. Persistent CSF rhinorrhea is potentially lethal. As it may lead to meningitis or brain abscess, surgical treatment is often required. Since St Clair Thompson's first description of 20 cases of nontraumatic rhinoliquorrhea in the late 1800s,<sup>3</sup> patients with a primary (spontaneous) CSF leak have presented a diagnostic and therapeutic challenge. Spontaneous CSF rhinorrhea has always been considered an infrequent entity that constitutes not more than 3% to 4% of all CSF leaks. A review article on spontaneous CSF leaks did not cite more than 10 cases.<sup>4</sup> In 1960, Troland<sup>5</sup> found only 33 cases of surgically treated primary CSF leaks mentioned in the world literature. Twenty-five years later, Hubbard et al<sup>6</sup> presented a series of 9 cases and in 1991, Tolley<sup>7</sup> reported 10 more cases. With 161 cases, Schick and coauthors<sup>8</sup> presented probably the largest series of surgically treated CSF leaks; of these 161 cases, 70 were caused by trauma, 36 by tumors, and 47 were complications of paranasal sinus surgery. Only 8 leaks in this series were spontaneous, caused by occult malformations of the skull base. These malformations are thought to be one of the main factors predisposing to spontaneous CSF leaks.<sup>9</sup> The second risk factor for spontaneous rhinoliquorrhea is obesity. Overweight increases intra-abdominal and intrathoracic pressure. This may affect blood circulation in cranial venous collectors and lead to the development of permanent benign intracranial hypertension.<sup>10,11</sup>

The most recent reports have demonstrated both an increasing prevalence of spontaneous CSF rhinoliquorrhea and a higher rate of spontaneous CSF leaks from fistulas of another etiology. In 2002, Dunn et al<sup>12</sup> reported that, in a series of 29 consecutive patients with CSF, rhinorrhoea was spontaneous in 10. Our personal experience also indicates that spontaneous CSF rhinorrhoea is not a condition as infrequent as was formerly believed. Of the 69 patients admitted to the hospital with a CSF leak during 3-year study period, 25 (36%) were diagnosed with spontaneous CSF rhinorrhoea.

It is essential to precisely locate the CSF fistula whenever surgical closure is considered. This is why the distribution of the sites of spontaneous CSF fistulas needs to be discussed. It was a common opinion that the cribriform plate and ethmoidal roof were the sites of the fistula in most patients with CSF, and the sphenoid sinus was rarely implicated in spontaneous CSF fistula. In contrast, our series, as well as some other recent reports,<sup>13,14</sup> shows that aside from innate skull base malformations, which usually occur in the cribriform plate, posterior and lateral walls of the sphenoid sinus are typical sites for spontaneous CSF fistulas. In our group of 21 patients operated on with the endonasal approach, a fistula of the sphenoid sinus wall was revealed in 9 cases. Recent studies<sup>13</sup> indicate that an overpneumatized sphenoid sinus, and particularly its lateral extensions, is

often a site where a CSF leak may appear. Extreme pneumatization of the sphenoid sinus may cause bone resorption in the skull base and, sometimes, a prolapse and tearing of the dura. A small meningocele may originate from bone dehiscence in the overpneumatized areas and this pathogenic mechanism, as well as the presence of an empty sella syndrome, should be considered important etiological factor for spontaneous CSF leak development.<sup>15</sup>

In our group extensive pneumatization of the sphenoid sinus was revealed in 6 patients, and a meningocele protruding through a bone defect in the lateral part of the overpneumatized sinus was found in 4 patients. It seems that with this particular type of CSF fistula, the presence of obesity and intracranial hypertension are not needed for the development of CSF rhinorrhea. Of the 21 patients, only 5 were obese; and in all patients with an overpneumatized sphenoid sinus, cerebrospinal pressure was normal.

Some cases of spontaneous CSF rhinorrhea may be due to an empty sella turcica. In this case, an intrasellar arachnoidocele can compress the pituitary gland against the posterior sphenoid sinus wall, erode the floor of the sella turcica, and lead to CSF rhinorrhea through the eroded sinus wall. The last 2 etiological factors may coexist. The combination of an extremely pneumatized sphenoid sinus and the empty sella syndrome theoretically increases the risk of developing CSF rhinorrhea. This can be illustrated by the case in our series where 2 small fistulas were found in the thin, eroded posterior wall of the sphenoid sinus.

Most of the recent publications dealing with endonasal surgical treatment of CSF rhinorrhea reported the efficacy of the method to be 85% to 95%,<sup>1,2,8,16</sup> but most of these publications dealt with all types of CSF leaks. Reports on endonasal surgical treatment of spontaneous leaks are still rare. However, spontaneous CSF rhinorrhea often presents the most challenging cases. Fistulas located in the lateral part of a large sphenoid sinus are extremely difficult to locate and repair with either the endoscopic endonasal or the transcranial approach. In our series, 20 patients were treated successfully and the only recurrence was a case where the fistula was in this particular location. The overall success rate was 95.2%.

#### CONCLUSIONS

Spontaneous CSF rhinorrhea is a clinical entity difficult to manage. Etiological factors of this disease may include obesity; innate skull base malformations; overpneumatized sphenoid sinus, particularly its lateral extensions; and empty sella syndrome. The possible role of these factors or their combinations must be considered when planning adequate therapy for a spontaneous CSF leak. All patients require a surgical intervention and the endonasal endoscopic approach appears to be the best option. As there is no universal technique, specific surgical options and choice of plastic materials depend on the fistula's size and location. However, treatment of CSF fistulas in the lateral part of the sphenoid sinus needs further perfecting.

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Corresponding author and reprints: Andrey S. Lopatin MD, Department of Ear, Nose, and Throat, Central Hospital, Presidential Medical Center, PO Box 4, 121614, Moscow, Russia (e-mail: asl@cch.pmc.ru).

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