*The Journal of Laryngology & Otology (2011)*, ***125***, *1185–1188.*

doi:10.1017/S0022215111001940

**CLINICAL RECORD**

# Bilateral, spontaneous cerebrospinal fluid rhinorrhoea: endoscopic, uninasal, trans-septal approach for simultaneous closure

M KURIEN[1], G A MATHEW[1], S L ABRAHAM[1], A IRODI[2]

*Departments of [1]ENT, Head and Neck Surgery, Speech and Hearing, and [2]Radiodiagnosis, Christian Medical College, Vellore, India*

## Abstract

*Background*: Bilateral, spontaneous cerebrospinal fluid rhinorrhoea is extremely rare, with only one previous case report (this patient developed contralateral cerebrospinal fluid leakage four years after successful endoscopic repair). We present the first English-language report of simultaneous, bilateral, spontaneous cerebrospinal fluid rhinorrhoea.

*Objective*: To recommend a simple alternative endoscopic technique for simultaneous closure of bilateral, spontaneous cerebrospinal fluid rhinorrhoea.

*Case report*: A 47-year-old woman presented with recent onset of bilateral, spontaneous cerebrospinal fluid rhinorrhoea, a recent history suggestive of meningitis, and a past history of pneumococcal meningitis. Bony defects on both sides of the cribriform plate were closed endoscopically in the same anaesthetic session, via a uninasal, trans-septal approach, enabling both leakage sites to be sealed simultaneously.

*Conclusion*: In cases of bilateral, spontaneous cerebrospinal fluid rhinorrhoea, uninasal, trans-septal endoscopic repair is a simple and effective technique for simultaneous closure of cerebrospinal fluid leakage.

**Key words:** Cerebrospinal Fluid Rhinorrhoea, Spontaneous; Nasal Cavity; Minimally Invasive Surgical Procedure

## Introduction

Cerebrospinal fluid (CSF) rhinorrhoea is the leakage of CSF from the arachnoid space into the nasal cavity via a defect in the dura, bone and mucosa.

Historically, the most common cause of CSF rhinorrhoea has been traumatic head injury.[1,2] Few authors[3,4] have reported iatrogenic injury (i.e. from functional endoscopic sinus surgery and neurosurgical procedures) as a common cause of CSF rhinorrhoea. Spontaneous CSF leakage is even less common; it has been variously reported to account for 25 per cent,[3] 40 per cent[4] and 49 per cent[2] of CSF rhinorrhoea cases.

In patients with untreated CSF rhinorrhoea, the incidence of developing one or more episodes of meningitis has been variously cited as 24 per cent,[4] 40 per cent[1] and 60 per cent of cases.[2,5]

Wigand first reported endoscopic repair of CSF rhinorrhoea in 1981, and it has now become the preferred treatment.[6]

In 2000,[7] Ramsden *et al.* reported bilateral, spontaneous CSF rhinorrhoea in a 47-year-old woman who presented initially with a CSF leak on the left side. Four years after successful endoscopic closure of this leak, she developed CSF rhinorrhoea on the right side; this was also repaired endoscopically, with no further episodes of leakage.

We present the case of a 47-year-old woman with bilateral, spontaneous CSF rhinorrhoea occurring simultaneously. This patient's CSF leaks were closed simultaneously using a uninasal, trans-septal approach, a previously unreported technique.

## Case report

A 47-year-old woman presented to the emergency department with a one-day history of fever, vomiting, headache and altered sensorium. She also complained of intermittent, bilateral, watery nasal discharge which increased on bending forwards, for the previous six weeks. She was a known hypertensive and had received medication for this for the past five years. She also gave a history of pneumococcal meningitis five years earlier, treated at our institution; at that time, she did not have any nasal symptoms or evidence of para-meningeal foci of infection.

Accepted for publication 21 February 2011 First published online 3 August 2011

Clinical examination revealed signs of meningeal irritation. Cerebrospinal fluid analysis was consistent with acute bacterial meningitis.

The patient was commenced on intravenous antibiotics (ceftriaxone) along with a short course of steroids.

Nasal endoscopy showed active leakage medial to the middle turbinate, in both nasal cavities. Fluid collected from both sides was subjected to biochemical analysis for sugar and protein; results were suggestive of CSF.

A high resolution computed tomography (CT) scan showed a small defect in the right cribriform plate (Figure 1). Magnetic resonance imaging (MRI) revealed a small meningocoele defect and adjacent CSF leakage via the right cribriform plate (Figure 2). However, no defect or leakage could be radiologically identified on the left side.

The patient was scheduled to undergo endoscopic repair of her CSF fistulae on completion of her anti-meningitic medication regime (i.e. two weeks of intravenous antibiotics as an in-patient, followed by oral antibiotics for one week as an out-patient). During her in-patient admission, she was also given the pneumococcal vaccine.

However, while taking oral antibiotics and awaiting surgery, the patient presented once again to the emergency department with signs of meningitis. She was admitted and treated for meningitis with intravenous ceftriaxone.

On the 14th day of her anti-meningitic regime, the patient underwent uninasal, trans-septal, endoscopic repair of her bilateral CSF leak. Following harvesting of fascia lata and fat from the right thigh, the fistulae were closed using standard endoscopic sinus surgery equipment, without the use of computer-assisted surgical navigation.

Initially, a right anterior ethmoidectomy was performed, followed by removal of the right middle turbinate to expose the roof of the right nasal cavity. Active CSF leakage was noted. The mucosa over the adjoining roof and superior part of the septum was elevated, revealing a large (4–5 mm) defect in the right cribriform plate area. The bony edges of the defect were identified.

Through the right nasal cavity, the superior part of the perpendicular plate of the ethmoid and adjoining mucosa were removed to expose the roof of the left nasal cavity, where active CSF leakage was noted. The mucosa around the leak was elevated to expose bare bone. A large (approximately 6–7 mm) defect was noted in the left cribriform plate area (Figure 3).

Fat was then tucked in between the dura and the bony defect on both sides, followed by fibrin glue and fascia lata grafting over the defect, including the bare bone on the roof of both nasal cavities (Figure 4). Turbinate mucosa plus fibrin glue was then applied, followed by Surgicel® and Gelfoam®. Both nasal cavities were then packed with bismuth iodoform paraffin paste.

Post-operatively, the patient was restricted to complete bed rest, with continuous lumbar subarachnoid drainage, for five days. Intravenous ceftriaxone was continued for a further eight days. The nasal pack was removed on the seventh post-operative day, following which the patient was discharged with a further week's course of oral antibiotics (cefixime).

At eight months' follow up, the patient had no evidence of recurrence of either CSF leakage or meningitis.

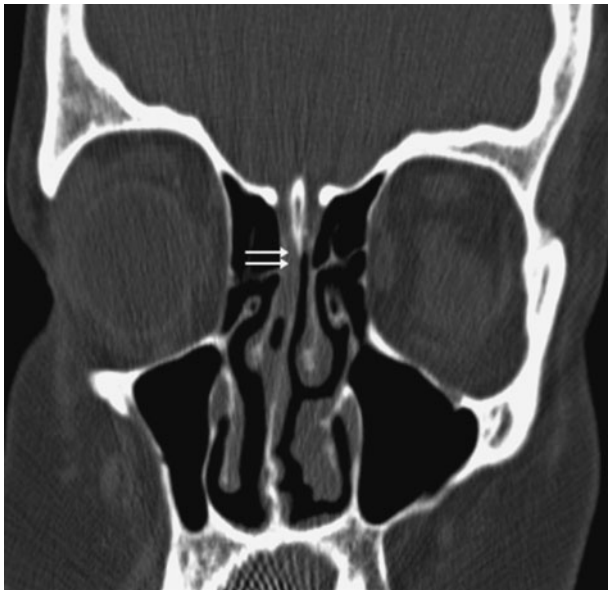

FIG. 1

Coronal, thin section computed tomography scan through the anterior cranial fossa, showing a small defect in the right cribriform plate, with a small, linear hypodensity extending from the anterior cranial fossa through the defect into the right nasal cavity (double arrows).

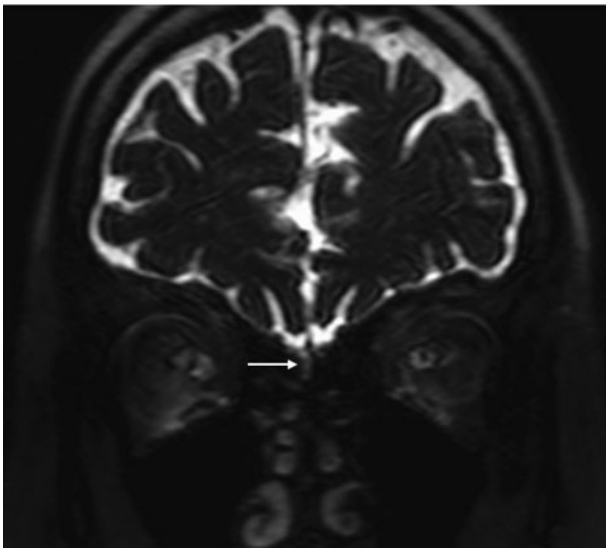

FIG. 2

Coronal magnetic resonance imaging scan, showing a small meningocoele and adjacent cerebrospinal fluid leak (arrow) through a defect in the right cribriform plate.

https://doi.org/10.1017/S0022215111001940 Published online by Cambridge University Press

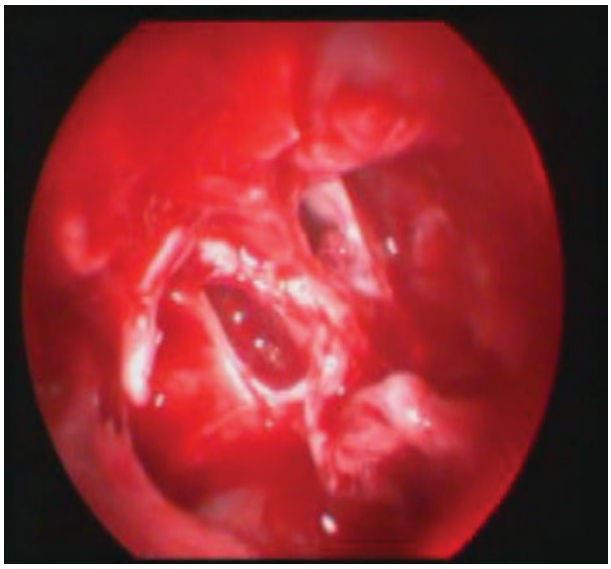

FIG. 3
Endoscopic view showing bilateral cribriform plate defects, with intervening superior aspect of the nasal septum, seen through the trans-septal surgical approach.

## Discussion

The first surgical intervention for repair of CSF rhinorrhoea was performed by Dandy in 1926, via an intracranial approach using a bifrontal craniotomy. In 1948, Dohlman was the first to use an extracranial approach via a nasofrontal incision. The first successful endoscopic closure of a CSF leak was performed by Wigand in 1981.[3,6]

The aetiology of CSF leaks can be broadly divided into traumatic (which can be subdivided into accidental and surgical) and non-traumatic (causes may be congenital, neoplastic or idiopathic). Traditionally, CSF rhinorrhoea caused by craniomaxillary trauma has been treated conservatively with bed rest, head elevation and strict sinus precautions,[4] as observations show that the majority of closed injuries resolve spontaneously. However, long term follow up of post-traumatic patients has found that 29 per cent subsequently develop meningitis.[5,8] Therefore, all post-traumatic, ongoing CSF leaks need to be repaired. Iatrogenic, post-surgical leaks should be closed as soon as they are identified. When there is no history of trauma (either accidental or surgical) and the CSF leak is spontaneous, the clinician should exclude benign intracranial hypertension (or pseudotumour cerebri), empty sella syndrome, morbid obesity and galactorrhoea.[2,4]

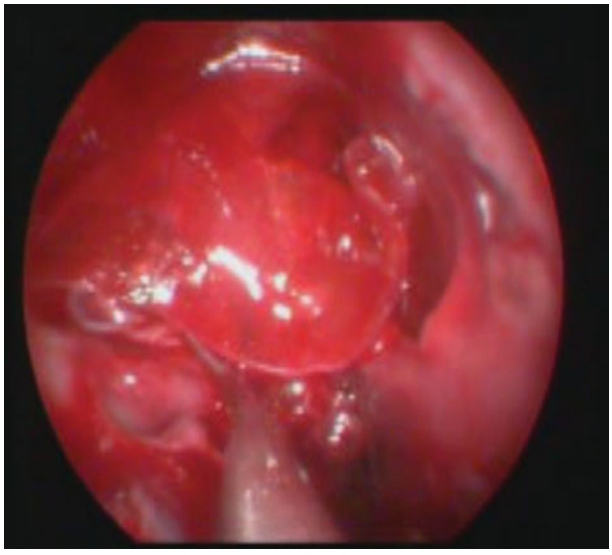

FIG. 4
Endoscopic view showing a single fascia lata graft applied over both defects.

In a suspected case of CSF rhinorrhoea, clinical examination should be followed by confirmation of the presence of CSF in the nasal fluid. Testing the fluid for the presence of β-2 transferrin remains the 'gold standard' for CSF identification.[3,4] Testing for β-trace protein has been performed by some authors, as it is highly sensitive, quicker (taking 20 minutes, versus 120 minutes for β-2 transferrin testing) and less expensive than β-2 transferrin testing. If there is any doubt over the results, β-2 transferrin testing can be performed subsequently.[3] Ye *et al.*[2] performed a retrospective study estimating the glucose concentration of nasal discharge, to confirm the presence of extracranial CSF. In our patient's case, we estimated the glucose concentration in the nasal fluid, as the other two tests were not available in our centre.

Clinic-based nasal endoscopy is of primary importance when attempting to identify the site of a CSF leak.

Additional pre-operative evaluation of the exact site of the leak is achieved using high resolution CT scanning of the paranasal sinuses and anterior skull base, this being accepted as the imaging modality of choice. Magnetic resonance imaging (with T2-weighted sequences) is indicated, in addition, when parenchymal or meningeal herniation is suspected.[3,4]

Many authors[4,9] have advised intra-operative localisation of the CSF leak using intrathecal fluorescein (after waiting approximately 30 minutes, a blue light is used to detect green-yellow fluid in the nasal cavity, indicating the leak site). Banks *et al.*[4] considered this to be the most accurate test to identify the CSF leak site. However, the use of intrathecal fluorescein is controversial due to its multiple reported complications.[10] Recently, Seth *et al.*[11] reported a series of 103 patients in which lack of intra-operative fluorescein visualisation did not exclude the presence of CSF leakage, as evidenced by a false negative rate of 26.2 per cent.

Presutti *et al.*[3] have advocated that the simple and safe Valsalva manoeuvre be performed intra-operatively, by the anaesthestist. This has the dual advantage of identifying the leak site and testing the adequacy of closure.

In our patient, the right leak location was confirmed pre-operatively using diagnostic endoscopy and high

https://doi.org/10.1017/S0022215111001940 Published online by Cambridge University Press

resolution CT scan with MRI. The left leak site, although noted on nasal endoscopy, could not be exactly defined using high resolution CT with MRI; however, its location was confirmed intra-operatively using the Valsalva manoeuvre.

Materials utilised for CSF leak closure included fat, fascia (temporalis[12] and lata[13]), free mucoperichondrial graft from the nasal septum,[3] and free middle turbinate mucosal graft without bone.[2] Composite osteomucosal or chondromucosal flaps have also been advocated[12,13] for the repair of defects larger than 3–4 cm. Although management must be tailored according to the aetiology and site of the leak, precise localisation of the leak and adequate preparation of the defect site is mandatory. A flat surface must be created, to which the graft can be made to conform fully, in order for closure to be successful.[4] The multilayer reconstructive technique has been recommended by Ye *et al.*[2]

- **This is the first reported case of bilateral, spontaneous, simultaneous cerebrospinal fluid (CSF) rhinorrhoea**
- **History-taking, examination, nasal endoscopy, high resolution computed tomography (CT) and (if possible) biochemical confirmation are required to diagnose and localise the leak site**
- **In this case, high resolution CT and magnetic resonance imaging did not identify the leak site on one side**
- **An endoscopic, uninasal, trans-septal approach (a new technique) achieved effective closure**

Presutti *et al.*[3] conducted a five-year, retrospective study of 52 patients, with endoscopic closure of CSF leaks using septo-mucoperichondrial grafting, and without lumbar drainage or fluorescein testing; they reported a success rate of 88.5 per cent for the first closure attempt. Banks *et al.*[4] undertook a 21-year, retrospective study of 193 patients, using endoscopic closure with intrathecal fluorescein localisation of the leak site and lumbar drainage in 73 per cent; they had an initial success rate of 85–90 per cent, and an overall success rate of 98 per cent. Ye *et al.*[2] conducted a 10-year, retrospective study of 69 patients, using endoscopic multilayer reconstruction without pre-operative fluorescein injection, and reported a success rate of 89 per cent at the first closure attempt.

## Conclusion

Non-traumatic, bilateral, simultaneous CSF rhinorrhoea is indeed a diagnostic and therapeutic challenge. Our patient represents the first reported case. An endoscopic, uninasal, trans-septal approach is effective in achieving simultaneous closure of leaks on both sides.

### References


1 Park J-I, Strelzow W, Friedman WH. Current management of cerebrospinal fluid rhinorrhoea. *Laryngoscope* 1983;**93**:1294–300
2 Ye H, Zuo J, Zhao H, Liu S, An H, Liu Y. Endonasal endoscopic repair of cerebrospinal fluid rhinorrhea in a series of 69 patients. *Br J Neurosurg* 2010;**24**:244–8
3 Presutti L, Mattioli F, Villari D, Marchioni D, Alicandri-Ciufelli M. Transnasal endoscopic treatment of cerebrospinal fluid leak: 17 years' experience. *Acta Otorhinolaryngol Ital* 2009;**29**:191–6 [in English]
4 Banks CA, Palmer JN, Chiu AG, O'Malley BW Jr, Woodworth BA, Kennedy DW. Endoscopic closure of CSF rhinorrhea: 193 cases over 21 years. *Otolaryngol Head Neck Surg* 2009;**140**(6):826–33. Epub 2009 Feb 28.
5 Eljamel MS, Foy PM. Non-traumatic CSF fistulae: clinical history and management. *Br J Neurosurg* 1991;**5**:275–9
6 Wigand ME. Transnasal ethmoidectomy under endoscopic control. *Rhinology* 1981;**19**:7–15
7 Ramsden JD, Corbridge R, Bates G. Bilateral cerebrospinal fluid rhinorrhoea. *J Laryngol Otol* 2000;**114**:137–8
8 Bernal-Sprekelsen M, Bleda-Vazquez C, Carrau RL. Ascending meningitis secondary to traumatic cerebrospinal fluid leaks. *Am J Rhinol* 2000;**14**:257–9
9 Lund VJ, Savy L, Lloyd G, Howard D. Optimum imaging and diagnosis of cerebrospinal fluid rhinorrhoea. *J Laryngol Otol* 2000;**114**:988–92
10 Keerl R, Weber RK, Draf W, Wienke A, Schaefer SD. Use of sodium fluorescein solution for detection of cerebrospinal fluid fistula: an analysis of 420 administrations and reported complications in Europe and the United States. *Laryngoscope* 2004;**114**:266–72
11 Seth R, Rajasekaran K, Benninger MS, Batra PS. The utility of intrathecal fluorescein in cerebrospinal fluid leak repair. *Otolaryngol Head Neck Surg* 2010;**143**:626–32
12 Stankiewicz JA. Cerebrospinal fluid fistula and endoscopic sinus surgery. *Laryngoscope* 1991;**101**:250–6
13 Kelly TF, Stankiewicz JA, Chow JM, Origitano TC, Shea J. Endoscopic closure of postsurgical anterior cranial fossa cerebrospinal fluid leaks. *Neurosurgery* 1996;**106**:1080–3



Address for correspondence:
Dr Mary Kurien,
Professor,
Dept. of ENT, Head and Neck Surgery,
Speech and Hearing,
Christian Medical College,
Vellore, Tamil Nadu, 632004, India

E-mail: ent2@cmcvellore.ac.in



Dr M Kurien takes responsibility for the integrity of the content of the paper
Competing interests: None declared


https://doi.org/10.1017/S0022215111001940 Published online by Cambridge University Press