

Endoscopic Repair of Spontaneous Cerebro-Spinal Fluid Rhinorrhoea: A Report of 3 Cases

B S Gendeh, MS(ORL-HNS)*, P J Wormald, FRACS*, M Forer, FRACS****, B S Goh, MD*, K Misiran, FFARCS****

*Departments of Otorhinolaryngology and **Anaesthesiology, Faculty of Medicine, National University of Malaysia Hospital (HUKM), Kuala Lumpur, ***Departments of Otolaryngology, Adelaide University, Adelaide and ****Royal North Shore Hospital, Sydney**, Australia

Summary

Three cases of spontaneous Cerebrospinal Fluid (CSF) rhinorrhea were managed at the National University Hospital, Kuala Lumpur. Case 1 had bilateral leak secondary to empty sella syndrome and the rest two cases had unilateral leak. Four transnasal endoscopic approaches were performed on these three cases since March 1999. The role of intrathecal Sodium Fluorescein is highlighted in localising the CSF fistula. Case 3 required postoperative lumbar drain as an adjunct. No recurrent leak was noted on post operative follow up in Case 2 and 3 ranging from nine to thirty two months. A recurrent left leak at six months was noted in Case 1 which could likely be due to her sudden bout of cough attacks and patient refused further surgical intervention.

Key Words: Spontaneous CSF Rhinorrhea, Intrathecal Sodium Fluorescein, Endoscopic closure

Introduction

CSF rhinorrhea involves a communication between the subarachnoid space and the nose. CSF rhinorrhea can be classified as either traumatic or spontaneous. The significance of CSF rhinorrhea is the potential development of fatal purulent meningitis via ascending infection through a persistent dural fistula, which mandates surgical repair.

CSF rhinorrhea is classified into traumatic and non-traumatic (spontaneous). The spontaneous

type is subdivided into primary where no underlying cause can be found or secondary to intracranial pathology (high or normal pressure). High pressure leaks include tumours, benign intracranial hypertension and hydrocephalus. Normal pressure leaks are due to empty sella syndrome, tumours, congenital defects, infection, arachnoid granulations, meningoencephaloceles and idiopathic conditions. Majority of CSF rhinorrhea are secondary to head trauma with an associated skull base fracture with only 3 to 4% considered spontaneous. Majority of

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Corresponding Author: Balwant Singh Gendeh, Department of Otorhinolaryngology, Faculty of Medicine, Hospital University Kebangsaan Malaysia, Jalan Yaacob Latif, Bandar Tun Razak, 56000 Kuala Lumpur

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spontaneous cases occur in adults in forth decade of life with females out numbering males (2:1). The initial onset is insidious, often mistaken for a feature of rhinitis. The site of fistulous communication in spontaneous CSF leak involves the dehiscence in the cribriform plate, fovea ethmoidalis, sphenoid sinus and rarely the frontal sinus.

The treatment of spontaneous CSF leak involves surgical closure of the defect. Traditionally spontaneous CSF leaks have been managed by the neurosurgeons via a frontal craniotomy with the success rate between 60 and 80%¹. It has been associated with significant morbidity such as frontal lobe retraction and anosmia. Wigand (1981) and Stankiewicz (1987) were the first to describe the transnasal endoscopic closure. In recent years it has gained popularity and is now considered by most otolaryngologist to be the treatment of choice for CSF rhinorrhea. Intrathecal Sodium Fluorescein allows precise preoperative endoscopic identification of CSF leak. Endoscopic approach has a success rate of between 76% and 95%². The morbidity of this approach is minimal, the sense of smell almost always preserved and there is a short hospital stay.

Three cases of spontaneous CSF rhinorrhoea closure by transnasal endoscopic repair seen over a 3 year period from January 1999 to December 2001 are presented.

Case 1

A 48-years Malay obese house-wife presented with a seven month history of intermittent bilateral clear nasal discharge (right more than left), worsen by straining or coughing. There was history of hyposmia and salty sensation in the throat. She had hypertension, diabetes and asthma. The CT scan was confirmatory of empty sella syndrome. There was no previous history of

head trauma, infection or nasal surgery. The nasal endoscopy examination was unremarkable. Upon leaning forwards, scanty amounts of clear fluid was collected from the right nostril. The presence of glucose in the clear fluid by laboratory test was suggestive of CSF.

CT cisternogram (CTC) was unremarkable. She was clinically and biologically eupituitarism. An endoscopic repair of the right CSF fistula was performed in March 1999. Prior to administering general anaesthesia, lumbar drainage was established and 0.1ml of 10% pure Sodium Fluorescein mixed with 10ml of CSF was administered intrathecally over 5 minutes. She was placed in Trendenberg prone position for about an hour. CSF leak was confirmed endoscopically on noticing a bright yellow green fluid draining from the right cribriform plate. There was no evidence of Sodium Fluorescein leak from the left nasal cavity. Her leakage was repaired using neural patch, histoacryl and merogel. The postoperative recovery was uneventful. Three months later she presented with intermittent watery rhinorrhea from the nonoperated nose. The nasal endoscopic examination was unremarkable. In March 2000, intrathecal Sodium Fluorescein examination was performed prior to general anaesthesia which revealed obvious CSF leak from left superior meatus. A right nasal endoscopic examination revealed a healed cribriform plate scar with no evidence of CSF leak. The site of leak was repaired using dural patch, histoacryl, middle turbinate free flap and merogel. The immediate post-operative recovery was unremarkable. At four months post-operatively, she complained of left watery rhinorrhea after bouts of coughing. Nasal endoscopy did not reveal an obvious CSF leak but the superior meatus appeared moist. On stooping, there was evidence of scanty clear fluid at the left nostril. The possible cause of the leak was the sudden bout of coughing resulting in raised intracranial pressure thus dislodging the weakly sealed fistulous plug. Except for the

occasional clear left rhinorrhoea on exertion, the patient was asymptomatic on follow-up at 24 months post-surgery. She refused further surgical intervention and was treated conservatively. She was advised to be on regular follow-up.

Case 2

A 47-years Malay house-wife was referred in June 2000 for right watery rhinorrhea of four months duration noted on straining. There was no previous history of head trauma, infection or nasal surgery. Nasal endoscopic examination was unremarkable. The clear fluid collected on stooping was sent for glucose analysis and was suggestive of CSF. CTC was unremarkable. In view of persistent history of clear watery discharge, she underwent endoscopic repair in July 2000. Intrathecal Sodium Fluorescein confirmed a CSF leak endoscopically arising from an arachnoid polyp at the right cribriform plate, medial to the middle turbinate. The arachnoid polyp was diathermised with bipolar and refashioned with microdebrider. The defect measured 3mm by 3mm with exposed dura. A mucoperiosteal flap was harvested from the opposite nasal septum and placed over the defect. Antero-inferior portion of the right middle turbinate was harvested as free graft to snugly fit in between the well placed mucoperiosteal flap, skull base and the remnant middle turbinate. Acrylic glue was applied over the snugly fitting middle turbinate free graft and covered with merogel. The postoperative recovery was uneventful. A follow-up at 18 months showed no evidence of recurrence.

Case 3

A 59-years Indian housewife was referred in August 2000 for right nasal spontaneous clear watery discharge. There was no history of head trauma, infection or nasal surgery. She was a known case of ischaemic heart disease,

hypertension and diabetes on regular medication. Nasoendoscopic examination revealed clear fluid pulsating through the right sphenoid ostium. CT scan revealed an opacified right sphenoid sinus with some evidence of bony erosion in the lateroinferior portion. She underwent endoscopic repair of CSF leak in June 2001. Intrathecal Sodium Fluorescein administration confirmed an obvious CSF leak endoscopically arising from a meningocele prolapsing through the lateroinferior wall. The meningocele was diathermised with bipolar and refashioned with microdebrider. The bony defect measured 3mm by 4mm and was closed with 'bath-plug' technique using fat harvested from the ear lobe. A free mucosal graft was harvested from the opposite middle turbinate and placed over the defect. A Valsalva maneuver was performed intraoperatively and confirmed no evidence of leak. A lumbar drain was inserted for 24 hours and then removed. The postoperative recovery was uneventful and the patient discharged on the third day. However, she had recurrent leak on stooping from the operated nose on post-operative day five and decided to get hospitalised on day ten. She denied history of exertion at home. Parenteral antibiotics were commenced immediately. After two days of conservative management with no evidence of clinical improvement, lumbar drainage was reinserted and removed after 72 hours with clinical recovery. She was discharged well and at follow up at 7 months there was no evidence of recurrence of CSF leak.

Discussion

Majority of CSF leaks are unilateral and only one bilateral CSF leak has been reported by Ramsden et al³. Case 1 is the second reported case of bilateral CSF leak in the literature.

Clear nasal discharge has been tested historically for glucose but is not diagnostic of CSF leak. False positive occurs due to the presence of lacrimal

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and nasal reducing substances. However, absence of glucose rules out CSF rhinorrhea. Beta-2 transferrin has become a diagnostic test for CSF fistula with a sensitivity nearing 100% and specificity of 95%. It is a protein found only in CSF, aqueous humor and perilymph.

Despite advances in imaging technology, the localization of CSF fistula can still be challenging. Precise preoperative localization of CSF leak is essential for surgical repair. The main problem of imaging modalities relates to the fact that spontaneous CSF leak may not be actively leaking at the time of the investigation as was obvious in all above three cases. High resolution CT scans have been advocated as the non-invasive radiological investigation of choice. The sensitivity rate has been reported from 50 to 100%. However, the main drawbacks are its high radiation to the lens and the bony defect may not correspond to the site of dural tear.

In the presence of frequent or constant CSF rhinorrhea, a CTC can be helpful in defining the exact site of leak. The overall accuracy improves from 40 to 92% if the leak is active. Since Case 1 and 2 were low pressure leaks, CTC was unremarkable. The disadvantages of CTC is it is invasive, time consuming requiring a lumbar puncture and intrathecal injection of contrast. The side effects include headache, seizure and infection⁴. In Case 3, endoscopic examination in the clinic was helpful in the diagnosis of CSF leak and therefore CTC was not performed.

The role of MRI in the evaluation of patients with CSF fistulae results from its superior demonstration of CSF and its continuity through dural fistulae with the paranasal sinuses or the middle ear. It is reserved for defining the nature of soft tissue namely inflammatory tissue, meningoencephalocele or tumour. It is noninvasive and has no radiation risk. However due to the inability of MRI to show bony defects CT scan remains a useful adjunct.

Preoperative intrathecal Sodium Fluorescein is helpful and diagnostic when imaging is not of much help. It was first used in 1960 by Kirshner and further developed by Messerklinger. Intrathecal Sodium Fluorescein is administered one to two hours prior to surgical procedure with the patient placed in the prone Trendelenberg position. Nasal endoscopic examination can precisely locate the site of the leak by illuminating the CSF light green or yellow. Complications of intrathecal Sodium Fluorescein include numbness and weakness of lower extremities, opisthotonus and seizures². It is rare and related to impurities and usage of high doses. Mattox and Kennedy² recommended 0.2mls while Wormald and McDonogh⁵ recommended 0.25mls of 5% Sodium Fluorescein could be used safely for the procedure. Kennedy feels that 0.1ml could be a safer dose to be used in smaller stature Asians.

In the first two cases, the localization of CSF fistulae was not clearly demonstrated by imaging modalities (CTC). In Case 3, the CT scan showed an opaque sphenoid on the affected nose, which was diagnostic on endoscopy in the clinic. Intrathecal Sodium Fluorescein was diagnostic in localizing the site of leak in all three cases.

The use of the endoscopes enables the surgeon to achieve good visualization of the site of the leak. Thus, careful removal of mucosa and fibrous tissue off the bone surrounding the defect is possible without significantly increasing the size of the defect. The application of the sealing tissue can then be performed under direct vision of the target area. The dissection is limited to the site of leakage with resultant low morbidity and high success rate. However, endoscopic repair is less suited for defects in the frontal sinuses with prominent lateral extension and defects greater than 3cm in diameter. For leaks from the supero-lateral wall of sphenoid may be approached via endoscopic route trans natural ostium of sphenoid but for leaks from infero-lateral wall as in Case 3, a lateral approach via an

ethmosphenodectomy is beneficial. A craniotomy repair may be preferred on failure of endoscopic approach.

There have been numerous reports describing various grafting techniques employed in endoscopic repair of CSF leak. The choice of the materials and techniques used during the endoscopic procedure depends on the experience and familiarity of the surgeon with various techniques. Weber et al (1996) used both free grafts and vascularised flaps with onlay and underlay techniques and successfully repaired all his 42 patients. Wormald and McDonough⁵ presented the 'bath plug' technique, which consisted of introducing a fat plug with vicryl suture into the intradural space in which the dural defects was less than 15mm. They claim that this technique would prevent high pressure from pushing the graft away from the defect. They also recommend the use of lumbar drain for a few days in an attempt to maintain a normal CSF pressure in patients who have choroids villi hypertrophy and are producing large amounts of CSF from ongoing longstanding chronic leakage. Valsalva maneuvers were performed to ensure the stability of the graft intraoperatively. Some authors feel that lumbar drains are unnecessary and have the added risk of sepsis and may allow air to be siphoned intracranially⁵. However others maintain that it is useful in patients with longstanding CSF leaks since it may lead to high CSF pressures being generated post-operatively and hence increase risk of failure².

In the above three cases, all were repaired endoscopically with various types of grafts used including allogenic material and vascularized flap, free flaps and 'bath plug' technique in Case 1, Case 2 and 3 respectively. Case 1 had a bilateral CSF leak secondary to empty sella syndrome which was repaired at one year apart and the leak recurred on the right after 3 months of surgery. Case 2 was a leak from cribriform plate secondary to arachnoid polyp which was successfully

repaired. Case 3 was a high pressure leak which recurred on the 5th postoperative day. This could have been avoided by placing a lumbar drain for more than 24 hours. According to Wormald and McDonough⁵, the lumbar drain should be left on free drainage 10cm above heart level with patient at 15 degree of head up to prevent air from being siphoned into the intrathecal space.

Whenever possible, the middle turbinate should be preserved to help stabilize the repair and optimize nasal function. Encephalocele if present is usually removed and the stump cauterized. The size of the defect dictates the type of material used in the repair. Larger defect (usually more than 3cm) may require bone or cartilage in addition to soft tissue. Placement of sealant (fibrin glue) is helpful followed by merogel (hyaluronic acid) which promotes healing and gets absorbed within 2 to 3 weeks. A nasal pack is not always necessary. Postoperative lumbar drain may be necessary in high pressure leaks or recurrent leaks. Patients are instructed to refrain from strenuous activity for 2 to 4 weeks. Avoidance of sneezing or coughing, use of stool softeners, elevation of the head and bedrest are essential to give the graft time to heal. The use of antibiotic is debatable. Close follow up is important to evaluate the success of the repair. Reported complications from endoscopic repairs include frontal lobe abscess and transient hemiparesis. Other complications include adhesion and septal perforation. No major complications were experienced.

Conclusion

Spontaneous CSF leak is very uncommon. It requires a high index of suspicion for diagnosis. A detailed history, endoscopic examination and appropriate investigations are important to confirm the diagnosis. A less invasive transnasal endoscopic technique has become the standard approach for CSF closure of less than 3cm with

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excellent initial success reported in the literature. The precise surgical technique may vary with equal success rate but highlights around clear endoscopic visualization and localization, proper selection of graft material and careful postoperative surveillance.

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