CASE REPORT

Idiopathic sphenoid sinus CSF rhinorrhoea

Manish Gupta,¹ Monica Gupta,² Gavinder Bindra,³ Sunder Singh⁴

¹Department of ENT, Gian Sagar Medical College, Chandigarh, Patiala, Punjab, India

²Department of Medicine, Government Medical College and Hospital, Chandigarh, India

³Department of Surgery, Gian Sagar Medical College, Patiala, Punjab, India

⁴Department of ENT, Gian Sagar Medical College, Patiala, Punjab, India

Correspondence to

Dr Monica Gupta,
monicamanish2001@gmail.com

SUMMARY

Cerebrospinal fluid (CSF) rhinorrhoea results from a direct communication between the CSF-containing subarachnoid space and the mucosa-lined space of the nasal cavity and paranasal sinuses. We present a case of 40-year-old woman, presenting with clear, watery discharge through the right nostril spontaneously. The CT cisternography confirmed the diagnosis of sphenoid sinus CSF rhinorrhoea, with no intracranial pathology. The patient was managed by transnasal endoscopic procedure, wherein bath plug technique was followed using temporalis fascia and overlay grafting with fascia lata and fibrin glue. The patient has been symptom free for the last year.

BACKGROUND

The spontaneous CSF rhinorrhoea is rare, and more so that from the sphenoid sinus.

CASE PRESENTATION

A 40-year-old woman presented to our otorhinolaryngology specialty with right nasal clear watery discharge of 6-month duration, aggravated by straining or bending forward. In the last month she had developed a headache which was gradually progressive; however, it was not associated with fever, vomiting, neck stiffness or blurring of vision. There was no history of trauma, nasal bleed or mucopurulent discharge. There was no history of recurrent sneezing, nasal itching or orofacial pain.

After examination the nasal mucosa on the right side was glistening and wet. The patient was seen pouring watery discharge from the right nasal cavity intermittently on bending forward, which was collected and sent for biochemical analysis. After diagnostic nasal endoscopy, the watery discharge was seen coming into the nasopharynx from the right sphenoid sinus ostia. The ophthalmological examination did not reveal any extraocular muscle weakness or papilloedema. The patient was subjected to bedside tests for detecting cerebrospinal fluid (CSF) rhinorrhoea. To perform reservoir sign, the patient was asked to immediately rise from the bed and place the chin over her chest. She was advised to stay in that position for one full minute. Clear fluid (CSF) was observed dripping from the right nose. The Double ring sign or the Target sign was negative suggesting that the CSF leak was not traumatic or associated with bleeding. Rest of the systemic examination was within normal limits.

INVESTIGATIONS

The initial investigations, haematological, biochemical and urine analysis were within normal limits. The nasal discharge sample glucose was 67 mg/dl

(normal range 50–80 mg/dl), protein was 20 mg/dl and no cells could be identified on cytology. The most sensitive laboratory test, that is, β_2 transferrin in CSF was positive in the discharge sample (absent in normal nasal secretions). Computed tomogram–cisternography with injection of non-ionic contrast (Omnipaque) into thecal space by lumbar puncture was done in the present case to localise the CSF leak. CT cisternography revealed well-defined lytic lesion 4 mm×4 mm in posterior wall of the sphenoid sinus (figures 1 and 2). The contrast was seen extravasating from the thecal space through this lytic lesion filling the right sphenoid sinus. Another similar lytic area 6 mm×7 mm was noted in skull vault, with both tables intact. Extensive pneumocephalus was seen (figure 3). Investigating for the cause of the lytic area in the skull bone, the peripheral blood film did not show increased plasma cells. Bone marrow aspiration was suggestive of a normocellular, normoblastic marrow with no evidence of multiple myeloma. The serum electrophoresis did not reveal 'M' spike. Urine was negative for Bence Jones proteins. The serum calcium, phosphorus and intact parathyroid hormone were all normal.

DIFFERENTIAL DIAGNOSIS

Accurate history can rule out a variety of other causes of clear watery nasal discharge. CSF otorrhoea presents as CSF rhinorrhoea after passage of fluid through eustachian tube. Sinonasal saline irrigations done by the patient as part of yoga exercise may present with similar clear watery discharge.

Figure 1 Sagittal section through right sphenoid sinus in CT cisternography, showing dye in pre-pontine cistern coming in the sphenoid sinus through defect in posterior bony wall (white arrow). Only posterior table seems to be involved due to obliquity of the defect.

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Figure 2 Adjacent sagittal section showing same defect (white arrow) involving the anterior table of the posterior wall of the sphenoid sinus.

Seasonal or perennial allergic rhinitis and vasomotor rhinitis may present with similar complaints but differentiated on detailed history and examination.

TREATMENT

The patient was started on conservative treatment with bed rest with 30° elevation of the head end. She was prescribed stool softeners, analgesics for headache and acetazolamide 250 mg four times a day. Prophylactic intravenous antibiotics, ceftriaxone and gentamicin were also started preoperatively to prevent meningitis. The patient was advised to avoid coughing, sneezing, nose blowing and straining.

The definitive procedure carried out in this case was extracranial endoscopic CSF fistula repair. Right-side uncinectomy, anterior-posterior ethmoidectomy and sphenoidotomy were done, fistula site was identified and leak confirmed on hyperventilation (figure 4). Surrounding mucosa was scraped off.

Figure 3 Axial cut CT showing air in brain and a 6 mm×7 mm lytic area in skull bone, with intact outer and inner lamina.

Temporalis fascia was tucked in the bony defect, fibrin glue and layer of fascia lata was placed over it in two layers (figure 5). Gelfoam and medicated ribbon gauze anterior packing were applied in the right middle meatus and the nasal cavity.

OUTCOME AND FOLLOW-UP

In the postoperative care the patient was kept on strict bed-rest for several days and given antistaphylococcal antibiotics. Acetazolamide was continued for a week. The nasal packing was removed after 2 days. The operative site was checked through serial nasal endoscopies and no postoperative leak was seen. The patient was advised to try to avoid strenuous activity, sneezing and coughing for 6 weeks after repair. The patient is under close follow-up, and is symptom-free for the last year.

DISCUSSION

The most common cause of CSF rhinorrhoea is trauma. In total, 80% cases are accidental and 16% attributable to iatrogenic trauma, due to intracranial or rhinology procedures like sinus surgery.¹ Non-traumatic, that is, spontaneous CSF rhinorrhoea accounts for only 4% of cases. Among the non-traumatic causes, majority are due to high intracranial pressure, as a result of brain tumour or hydrocephalus while normal pressure CSF rhinorrhoea is seen in cases of congenital dehiscence, osteomyelitic erosion of the skull base or focal atrophy brain (olfactory or sellar).¹ Idiopathic cause is the rarest and often a diagnosis of exclusion.

Spontaneous CSF leaks have been estimated as infrequent entity, but recently this idea has changed.² Skull base malformations and obesity are considered risk factors for their development.³ As in our case the search for the cause of lytic lesion, we ruled out various tumours which metastasize to bone-like breast, lung, renal, ovarian and pancreatic cancer and multiple myeloma. They often present with severe bone pain, pathological fractures, leucoerythroblastic anaemia, hypercalcaemia and nerve compression syndromes. The lytic lesion may also arise due to rise in osteoclast-activating factors like parathyroid hormone-related peptide. Moreover, because of increase in bone formation in patients with osteolytic lesions, the serum alkaline phosphatase levels are increased and there is an increased uptake on bone scan at the site of lesion. In this case all these parameters were within normal limits, thus this lytic lesion was in all likelihood idiopathic.

Figure 4 Endoscopic picture showing posterior wall of sphenoid sinus with bony defect and raw bone (black arrow) around after removal of surrounding mucosa.

Figure 5 Endoscopic picture showing ball probe pushing temporalis fascia in the defect.

The diagnosis of CSF rhinorrhoea is both clinical and radiological. The presence of CSF in clear nasal discharge should be established by the analysis for CSF markers. Localisation of the leak site involves diagnostic nasal endoscopy and radiological investigation, like high-resolution computerised tomography with intrathecal administration of non-ionic contrast (CT cisternography) and flow sensitive MRI. The leak must be active at the time of the scan for visualising the site. Intrathecal fluorescein can also be used intraoperatively to detect the site of leak in the nasal cavity with a 490 nm light generated by an optical filter.

Besides controlling the leak, the main aim of the closure of CSF rhinorrhoea is to prevent complications like ascending meningitis, pneumocephalus and postmeningitic hydrocephalus.⁴ The operative intervention is advised when the leak is post-traumatic and persistent after 4–6 weeks of conservative treatment, intraoperative recognition of a leak (during sinus surgery, skull base surgery and craniotomy), large defects/leaks (especially in association with pneumocephalus), all cases of spontaneous CSF fistula, delayed post-traumatic leak and CSF leak with a history of meningitis.⁵ The graft material used depends mainly on the surgeon's experience and did not significantly influence the success rate.⁵ The various autograft-free or pedicled may be used like—temporalis fascia, fascia lata, muscle plugs, pedicled middle turbinate flaps (mucosa alone or mucosa and bone), autogenous fat, free cartilage grafts (from the nasal septum or the cartilaginous auricle) and free bone grafts (from the nasal septum or calvarium).

The efficacy and safety of the trans-sphenoidal approach for the resection of pituitary tumours and the treatment of associated CSF rhinorrhoea has encouraged surgeons to use the transnasal route for the repair of all CSF leaks in the middle cranial fossa and sphenoidal sinus region.⁶ The transnasal approaches to the sphenoid sinus have been proven to be safe and effective in the treatment of CSF rhinorrhoea associated with post-trans-sphenoidal pituitary surgery, postoperative anterior cranial base surgery and even medial intrasphenoidal and sellar encephalocele.^{3,6}

The operative microscope provides direct illumination and magnification of the operative field. However, it allows a detailed view only of superficially located structures and requires manipulation and retraction of intact superficial structures, leading to iatrogenic trauma.⁷ The use of endoscope avoids this trauma to superficial normal structures, giving a

good view of the surgical field and also avoids trauma associated with transcranial approaches.

The traditional approaches like frontal craniotomy carry various advantages like the direct inspection of adjacent brain tissue and visualisation of the dural defect. The repair can also be done even under conditions of increased intracranial tension and even if efforts to localise the leak fail, blind repair is possible in this approach. However, it has many disadvantages like brain handling resulting in oedema, sagittal sinus injury, anosmia, intracranial haemorrhage, seizures, memory deficiencies and behavioural disorders, besides high failure rates of 25%.

In the transnasal endoscopic plugging, the main steps are accurate localisation of the defect, creation of a raw surface around the defect (to accept the graft and help in the formation of synechiae to support the seal later), plugging of the defect with fat covered with fascia lata supported by absorbable gelatin and merocel. The differences between various surgeons lie in the use of fluorescein to locate the defect, the plugging materials and the technique of underlay or overlay grafting, the use of fibrin glue and the need for lumbar drainage postoperatively.⁵

The transnasal endoscopic procedure provides excellent visualisation and precise localisation of the defect in the ethmoid roof, cribriform plate and most of the sphenoid sinus. It also ensures accurate positioning of the graft material over the defect. It is well tolerated, carries less morbidity and short inpatient stay and accelerating return to work, therefore cutting indirect costs as compared with intracranial repair.⁵ In children, it reduces the impact on the still-developing splanchnocranium.

Endoscopic repair of skull base defects is successful in over 90% of cases.⁸ The factors responsible for failure of skull base repair techniques or need for secondary repair are inability to localise the defect, development of new defect, prior sinus or skull base surgery or craniotomy, prior radiation therapy and intracranial infection.⁸

Learning points

- ▶ Most cases of cerebrospinal fluid (CSF) rhinorrhoea are traumatic and some of these may be iatrogenic, especially neurosurgical or rhinology procedures like sinus surgery.
- ▶ Traumatic cases generally respond well to conservative management. However, non-traumatic CSF rhinorrhoea require operative repair and extracranial techniques are preferred line of management.
- ▶ The transnasal endoscopic procedure precisely localises the defect, ensures accurate graft positioning, is well tolerated and carries less morbidity.

Contributors MaG and MoG are the physician and the ENT specialist, respectively, who have seen and managed the case and have drafted the initial version of this manuscript. GB and SS have assisted during the patient's workup, surgery and postoperative management. All the authors have critically analysed the text, images and contributed significantly in shaping the final manuscript.

Competing interests None.

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