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Cerebrospinal Fluid Rhinorrhea with Encephalocele Post Septoplasty

Abstract - Septoplasty is a common surgical operation done for patients with deviated septum. Cerebrospinal fluid (CSF) leak post septoplasty is one of the rare complications and delayed diagnosis can lead to herniation of brain substance through the skull base defect and lead to encephalocele. We reported a case of CSF rhinorrhoea with encephalocele complicating septoplasty which was repaired surgically. We discuss the clinical importance of anatomical variations and causative mechanisms of CSF rhinorrhoea, and the diagnosis and management of the case. Early diagnosis and management of the CSF leak with encephalocele post-septoplasty are important to prevent life threatening events and for better outcome.

***Keywords*- Cerebrospinal fluid leak; Encephalocele; Septoplasty; Complication; Repair**

1 INTRODUCTION

Septoplasty is a surgical operation to remove deviated nasal cartilage or bone to correct a nasal septal deformity. It is a common procedure but can lead to major complications like major skull base injuries with cerebrospinal fluid (CSF) leaks [1,2]. CSF leak with encephalocele post-septoplasty is extremely rare with an estimated incidence of 0.01% to 0.1% [1].

2 CASE REPORT

A 31-year-old Indian male, with underlying allergic rhinitis initially presented to a private hospital with the symptoms of bilateral nose block, nasal discharge, and occasional facial pain for years, with examination revealed a severely deviated nasal septum to the left and inferior turbinate hypertrophy. He then underwent septoturbinoplasty in that hospital and was discharged well.

Six months later, he presented to us with the symptoms of unilateral left nasal block and nasal clear discharge which worsened when he bent forward and aggravated during heavy lifting or straining. There was no vision abnormality, headache, facial pain, anosmia, epistaxis, or symptoms and signs of meningitis.Nasal endoscopy noted deviated nasal septum to the left, with left nasal cavity mass, medial to the left middle turbinate, likely dura as the mass was increased in size during a Valsalva maneuver (Figure 1).

Computed tomography (CT) of paranasal sinuses showed an anterior cranial fossa left cribriform plate bony defect with an encephalocele protruding through the defect into the superior aspect of the left nasal cavity medial to the left middle nasal turbinate. The defect measures 0.6cm. Mild to moderate polypoidal mucosal thickening at left maxillary sinus and minimal mucosal thickening at left posterior ethmoidal air cells, right sphenoid sinus, and right maxillary sinus (Figure 2).

The patient was then proceeded with left functional endoscopic sinus surgery, CSF leak repair, and reconstruction. The defect was repaired multilayer by periumbilical fat graft, left middle turbinate tissue, and tissue glue application (Figure 3). The operation went uneventfully and no further leak was seen 1 month postoperatively (Figure 4).

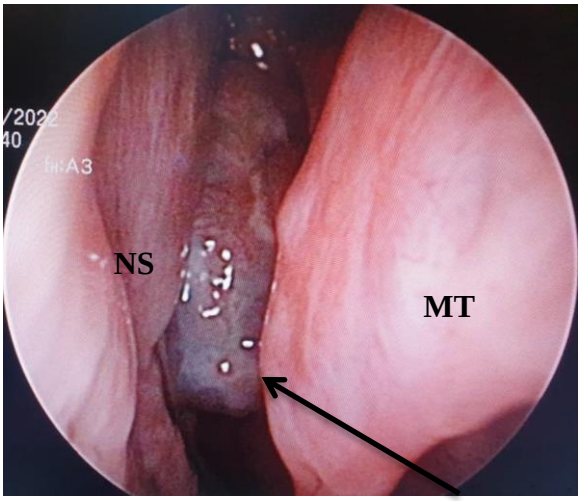

Figure 1: Endoscopic view of the nasal polypoid lesion (encephalocele sac) between nasal septum and left middle turbinate (arrow). MT: middle turbinate, NS: nasal septum.

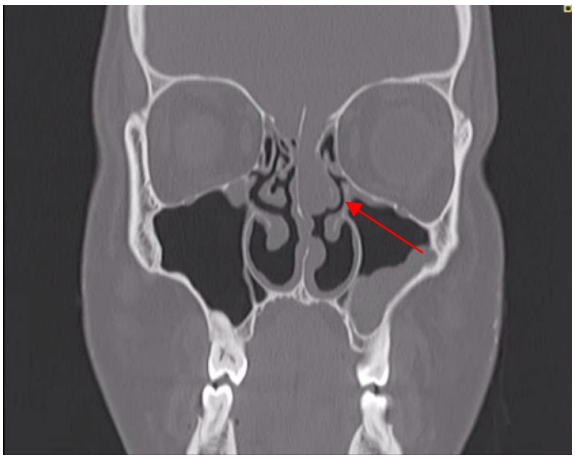

Figure 2A

Figure 2B

Figure 2: CT Paranasal sinus (A coronal) showing an encephalocele protruding the defect into the superior aspect of the left nasal cavity (arrow), (B sagittal) skull base defect is shown and measuring 0.6cm

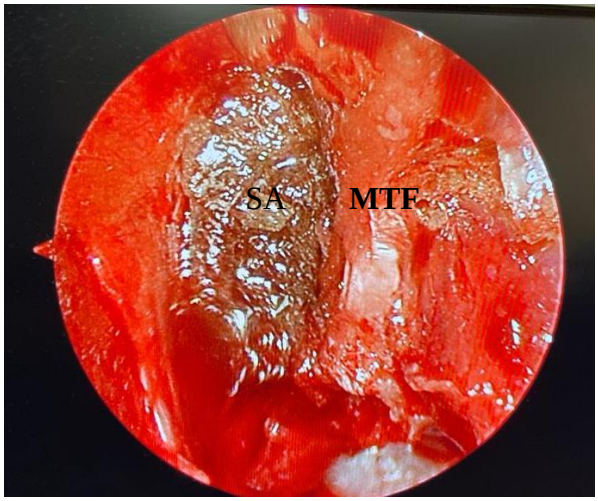

Figure 3: Intraoperative surgical repair of anterior skull base defect by multi-layer technique: Periumbilical fat graft and surgical adhesive (first layer), absorbable hemostat and surgical adhesive (second layer), left middle turbinate tissue and surgical adhesive (third layer).

SA: Surgical Adhesive; MTF: middle turbinate flap

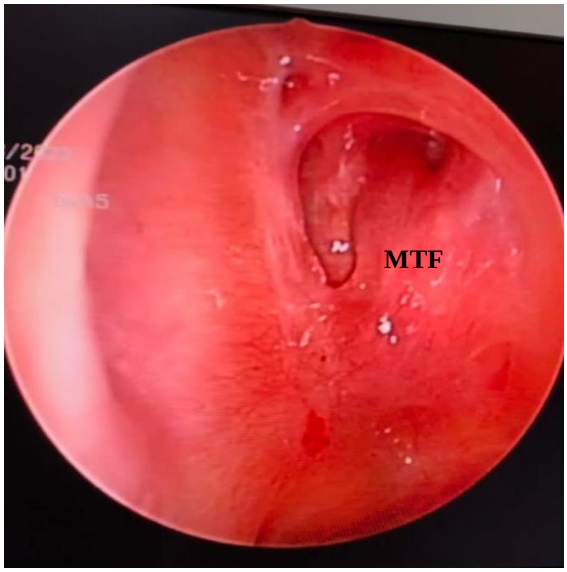

Figure 4: Post-operative 1 month showed healthy middle turbinate flap (MTF) in situ and no further CSF leak seen

3 DISCUSSION

CSF rhinorrhea is the result of abnormal communication between the subarachnoid space and the sinonasal cavity [3,4]. Whereas intranasal encephalocele is an uncommon lesion with the herniation of brain substance through the skull base defect [5]. Nasal encephaloceles are classified based on their location of defect [5]. It can be classified into frontoethmoidal and basal

encephaloceles [5]. Basal encephalocele can be further classified into transethmoidal, sphenoethmoidal, transsphenoidal, and fronto-sphenoidal [5]. The main symptom of encephalocele is CSF rhinorrhoea [5]. 80-90% of the cases are related to trauma, this being either a head injury or iatrogenic following endoscopic endonasal surgery [3]. In our case, this patient had a CSF leak with transethmoidal encephalocele post-septoplasty, which is extremely rare.

In such cases, patients might present with unilateral nasal symptoms and the lesion may be easily misdiagnosed as a nasal polyp [4]. A biopsy should not be done in such cases as it can lead to serious neurological complications like severe bleeding, neurological defects, seizures, or death [4]. Patients with a history of nasal surgery who presented with a unilateral intranasal mass should always raise the clinical suspicion of encephalocele, like in our case.

The most likely cause of CSF leak with encephalocele in this case will be iatrogenic damage to the cribriform plate. Mechanisms of injury include elevation of the septal mucoperichondrium with a Cottle elevator and tunneling too superiorly on the septum, beyond the limits of the ethmoid roof, or by fracturing the perpendicular lamina, which subsequently can fracture the cribriform plate [2,6,7]. These areas are at risk because of the connection to the dura by a weak and thin bone structure [2]. The anterior cranial fossa is the usual site of this complication, with the roof of the ethmoid sinus and the cribriform plate as the most common sites [2,6,7].

Early diagnosis is important as it can lead to enlargement and remodeling of the bone over time and a dural defect, leading to herniation of the meninges and brain tissue through the defect, as we can see from this case [2]. CSF rhinorrhea can eventually lead to life-threatening sequelae like meningitis, encephalitis, and cerebral abscess formation. High-resolution CT is the preferred method of localizing the site of the skull base defect but can be coupled with magnetic resonance imaging (MRI) or cisternography [8]. MRI gives details of the intracranial anatomy and pathology in multiple planes [8]. The only disadvantage is limited access, as in our case given the long waiting time during the pandemic season, and we did not want to delay the management for him. However, most of the literature shows that combining CT and MRI accurately localized the CSF leak and encephalocele [8].

The majority of traumatic CSF rhinorrhea can be managed conservatively for up to two weeks, and up to 85% of CSF leaks will heal spontaneously [3,9]. Conservative measures include bed rest, laxatives, and the avoidance of straining [3]. Conservative management was not recommended in this case as it is complicated by dural defects. Surgical intervention is recommended in the cases of persistent CSF rhinorrhoea, which is at risk of meningitis, and those with intracranial pathology [9].

Surgical repair includes an open approach or a transnasal endoscopic approach. Open approaches can be done in cases like large encephaloceles, extensive multiple defects, and leaks that are associated with intracranial lesions or hematomas [8,9]. It provides direct access to the defect and allows repair of multiple sites, but is associated with higher morbidity and recurrence as compared to endoscopic approaches [8]. As compared, endoscopic approaches bring excellent outcomes and reduce morbidity. Endoscopic endonasal approaches should be the preferred method of repair, with a greater than 90% success rate [8].

Identification and delineation of the bony defect at the skull base, bipolar diathermy of the encephalocele sac, resection of the neck of the meningoencephalocele sac, and multilayer closure of the resultant defect are the key steps in the surgical management of intranasal encephalocele or meningoencephalocele [10]. Bipolar cautery of the encephalocele provides a safe excision as the brain tissue may not be functioning [10]. However, if the brain tissues are sessile and their implantation base is broad, the brain content may be functional [10]. In our case, we reduced the encephalocele back through the skull base defect without bipolar cauterization.

A variety of graft materials can be used for the repair of the skull base, including fat, bone, allografts, free mucosal or fascial grafts, vascularized flaps as well as synthetic grafts and sealants to hold the repair in place. There is currently no evidence that supports one material being superior to another, and their use very much depends on the site and size of the defect as well as the surgeon's preference [8]. Small post-surgical defects can be repaired endoscopically with mucosal grafts, while a pedicled nasal septal flap can be used for large defects [8,9].

In our case, we used a multilayer technique to repair the skull base defect, as it is safer to hold the repair and can prevent recurrence. It was repaired with a periumbilical fat graft layered with absorbable hemostat and surgical adhesive as the first layer, followed by another layer of absorbable hemostat and surgical adhesive as the second

layer, then left middle turbinate tissue with another layer of surgical adhesive as the third layer. The nasoseptal Hadad-Bassagaisteguy flap is currently the gold standard for the closure of skull base defects after endoscopic transnasal surgery [8]. This flap is both reliable and robust due to the robust vascular pedicle [8]. However, it was attempted in this case but failed due to a deficiency of septal mucosa and limited flap tissue to reach and cover the skull base defect.

4 CONCLUSION

Septoplasty is one of the most widely performed operations. Although complications are rare but can be serious and life-threatening complication if not managed quickly and appropriately. Hence understanding the anatomic knowledge, mechanism of injury and preoperative detection of anatomical variations in the nose and skull base are essential when performing any sinonasal procedure for a better outcome.

CONFLICT OF INTEREST

The authors report no conflicts of interest. The authors are responsible for the content and writing of the paper.

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5 REFERENCES

[1] Youssef A, Ahmed S, Ibrahim AA, Daniel M, Abdelfattah HM, Morsi H. Traumatic cerebrospinal fluid leakage following septorhinoplasty. Archives of Plastic Surgery. 2018 ;45(4):379–83.

[2] Bloom JD, Kaplan SE, Bleier BS, Goldstein SA. Septoplasty Complications: Avoidance and Management. Otolaryngologic Clinics of North America. 2009 ;42(3):463–81.

[3] Abuabara A. Cerebrospinal fluid rhinorrhoea: diagnosis and management. Med Oral Patol Oral Cir Bucal. 2007;12(5):397-400.

[4] Aslier M, Kasapoglu F. Diagnosis Algorithm in Single-Sided Nasal Masses. European Journal of Rhinology and Allergy. 2019;2(3):82–5.

[5] Nesrin Gürsan, Mehmet Dumlu Aydin. Intranasal encephalocele: A case report. Turkish Journal of Medical Sciences. 2003;33:191–4.

[6] Leong AC, Patel TR, Rehman F, Oyarzabal M, Gluckman P. Cerebrospinal fluid rhinorrhea complicating septoplasty: A novel mechanism of injury. Sage Journal. 2010;89(1):27–9.

[7] Imre A, Pinar E, Ahmet Ata Ece. Ethmoidal meningoencephalocele and cerebrospinal fluid leak after septoplasty: a rare complication. ENT Updates. 2017;7(2):108–11.

[8] Thibaut Van Zele, Dewaele F. Traumatic CSF leaks of the anterior skull base. B-ENT. 2016;26(2):19–27.

[9] Le C, Strong EB, Luu Q. Management of anterior skull base cerebrospinal fluid leaks. J Neurol Surg B Skull Base. 2016;77(5):404–11.

[10] Ruggeri CS, Aragón GS, Acosta L, Figueroa E. Endoscopic management of intranasal meningoceles and meningoencephaloceles. Journal of Otolaryngology-ENT Research. 2019;11(3):175–9.