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Ethmoidal meningoencephalocele and cerebrospinal fluid leak after septoplasty: a rare complication

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Abstract

A 24-year-old man referred to our clinic with complaint of intermittent right-sided watery rhinorrhea. Patient underwent nasal septoplasty one year ago and rhinorrhea occurred two weeks after the surgery. Rhinorrhea was ignored and then patient developed meningitis two months after the surgery. Subsequently, meningoencephalocele formation developed. In this case report, we present a case of meningoencephalocele associated with cerebrospinal fluid leak diagnosed one year after the septoplasty. Septoplasty is usually regarded as a relatively safe operation. However, forceful maneuvers to perpendicular lamina of the ethmoid bone may cause breakdown of the skull base structures, particularly the horizontal lamella of the cribriform plate. When this occur, immediate management is necessary to prevent intracranial complications including meningitis, intracranial abscess, and pneumocephalus. Delay in the diagnosis of such injury may cause erosion of the bone and gradual herniation of the intracranial contents through the skull base defect.

Keywords: Cerebrospinal fluid leak, complication, endoscopic repair, meningoencephalocele, septoplasty.

Özet: Septoplasti sonrasında etmoidal meningoensefalosel ve serebrospinal sıvı kaçağı: Nadir görülen bir septoplasti komplikasyonu

Yirmi dört yaşında erkek hasta aralıklarla olan sağ taraflı su gibi burun akıntısı şikayeti ile kliniğimize refere edildi. Hasta 1 yıl önce septoplasti operasyonu olmuş ve operasyondan 2 hafta sonra rinore ortaya çıkmıştır. Rinore şikayeti ihmal edilen hastada ameliyattan 2 ay sonra da menenjit gelişmiştir. Bunu takiben de meningoensefalosel formasyonu oluşmuştur. Bu olgu sunumunda, septoplasti operasyonundan bir yıl sonra tanı konan meningoensefalosel ve eşlik eden serebrospinal sıvı kaçağı olgusunu sunduk. Septoplasti genellikle güvenli bir operasyon olarak kabul edilmektedir. Bununla birlikte, etmoid kemiğin perpendiküler laminasına zorlayıcı kuvvetli manevralar uygulanması kafa kaidesi yapılarında özellikle de kribriform plakanın horizontal laminasında fraktüre neden olabilmektedir. Böyle bir durumda, menenjit, intrakraniyal apse ve pnömosefalus gibi intrakraniyal komplikasyonların gelişmesini önlemek için hemen tamir edilmesi gerekmektedir. Böyle bir hasarda tanıda gecikme kafa kaidesindeki kemikte erozyona ve zamanla bu defektten intrakraniyal içeriğin herniye olmasına neden olabilmektedir.

Anahtar sözcükler: Endoskopik onarım, komplikasyon, meningoensefalosel, septoplasti, serebrospinal sıvı kaçağı.

Meningoencephalocele and cerebrospinal fluid (CSF) rhinorrhea of the anterior cranial fossa is a well known entity resulting from iatrogenic and non-iatrogenic causes including congenital malformations and skull base erosion from intracranial tumors. Iatrogenic CSF leaks frequently occur during endonasal transsphenoidal pituitary surgery and functional endoscopic sinus surgery.^[1,2]

Septoplasty, one of the most common operations performed in otolaryngology, is usually regarded as a relatively safe operation. However, various complications including CSF leak following septoplasty are reported previously.^[3,4] Meningoencephalocele formation associated with CSF leak following septoplasty is very rare.^[5]

In this case report, we present a case of meningoencephalocele associated with CSF leak diagnosed one year after nasal surgery. The probable pathological mechanism and endonasal endoscopic management of this complication were discussed.

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Case Report

A 24-year-old man was referred to our tertiary clinic with complaint of profuse, intermittent right-sided watery rhinorrhea. His medical history revealed that the patient underwent nasal septoplasty at his local hospital one year ago. As noted in the perioperative records, deviated perpendicular lamina of the ethmoid bone was taken out after fracturing with Brunings nasal forceps. On the postoperative 14th day control, watery right-sided rhinorrhea occurred but was initially ignored. Eight weeks later, patient developed meningitis with complaint of headache and clouding of consciousness. Lumbar puncture revealed purulent (1000 leucocyte/mm³) and highly viscous CSF. Patient was successfully treated and discharged from the hospital. However, intermittent rhinorrhea has continued since patient was discharged. Nasal endoscopy revealed a small polypoid lesion between nasal septum and middle turbinate (Fig. 1a). Valsalva maneuver elicited profuse right-sided watery rhinorrhea. Paranasal sinus computed tomography (CT) revealed an asymmetrical anterior skull base height which was fairly suspicious for a small right cribriform encephalocele and bony defect of the right olfactory cleft base. Subsequent fast imaging employing steady-state acquisition (FIESTA) magnetic resonance (MR) imaging confirmed

the cribriform lamina defect (5×5 mm) and meningoencephalocele sac in which a part of gyrus rectus herniated throughout the defect (Fig. 1b). The patient underwent endonasal endoscopic repair. The meningoencephalocele was cauterized using a bipolar device. The skull base around the defect site was circumferentially demucosalized and cauterized to prevent postoperative mucocele formation. The skull base defect was repaired using underlay septal cartilage graft, followed by an overlay septal mucosa graft (Fig. 2). The overlay graft was bolstered with pieces of Gelfoam. Bed rest was recommended and diuretics were prescribed. The patient's postoperative course was uneventful. At his first-year follow-up, no evidence of rhinorrhea was present.

Discussion

CSF leak and meningitis is a dreaded complication of rhinologic and neurosurgical procedures. Rhinorrhea is caused by a breakdown of the skull base and closely adherent dura causing connection between subarachnoid space and nose. Therefore, any case of rhinorrhea associated with a recent history of endonasal surgery warrants further investigation.^[6] Particularly, endoscopic sinus surgery and skull base surgery are due to potential for iatrogenic CSF leak. However, uncontrolled twisting manipulations of the perpendicular



Fig. 1. (a) Endoscopic view of nasal polypoid lesion (meningoencephalocele sac) between middle turbinate and nasal septum. M: middle turbinate, S: superior turbinate, NS: nasal septum. *Meningoencephalocele sac. (b) Meningoencephalocele sac was shown in FIESTA T2-weighted MR sagittal section.

lamina of the ethmoid bone during septoplasty may elicit skull base injury leading life-threatening complications.

Anterior skull base is extremely vulnerable to trauma because of the thin bone and attachment of dura to the bone. The perpendicuar crista galli, comparatively strong and thick bone complex, forms the midline structure. The ethmoid labyrinth and cribriform plate supports the olfactory bulbus and is punched by various foramina for the passing of the olfactory filaments and small arachnoid pouches. Fractures of the ethmoid bone related with damages to the arachnoid and olfactory ligaments. A few authors have reported CSF leakage after septoplasty.^[3,7,8] They proposed that these slit-shaped dehiscences at the horizontal lamella of the cribriform plate or perforation of the cribriform plate likely resulted from forceful operative technique applied for removal of the perpendicular plate of the ethmoid bone.

Thakar et al.^[9] reported two patients with delayed CSF leak which occurred three months after the septoplasty operation. They attributed this delayed leaks to following dural pulses at the injury site, leads to bone resorption and resultant dural herniation and rupture. Previously, Taveras and Ransohoff^{10]} suggested a similar system for the disposition of a leptomeningeal cyst: iatrogenic trauma produces a



Fig. 2. Intraoperative endoscopic view after skull base repair with underlay septal cartilage. MT: middle turbinate, NS: nasal septum, SB: demucosalized skull base bone. *Underlay septal cartilage.

skull fracture; meninges herniate through the defect and pulsation gradually pushes more meninges through the defect; growing sac subsequently corrodes the bone. In our current patient, at first year after the septal surgery, meningoencephalocele sac was observed in nasal cavity between the middle turbinate and nasal septum. This theory may explain the acquired meningoencephalocele in our patient.

Removing the deviated posterior bony septum by applying forceful maneuvers to perpendicular lamina of the ethmoid bone remains common practice in septoplasty. We agree with Thakar et al^[9] who proposed that sharp instruments such as bone scissors may be much safer than grasping forceps to fracture bony septum. Small amounts of the CSF leak might not be recognized. In addition, neurologic findings are not present straightway postoperatively, if there is a noteworthy rapidly growing hematoma or pneuomcephalus. When skull base injury occur, immediate management is necessary to prevent intracranial complications. The current management of CSF leak and meningoencephalocele are endoscopic endonasal repair which has evolved significantly during the past decade.^[11] Safety and efficacy of this approach have surpassed the traditional open approaches. In the current case we also performed a successful endonasal endoscopic repair with uneventful postoperative course.

In conclusion, although the CSF leak following nasal septoplasty is rare, forceful maneuvers to perpendicular lamina of the ethmoid bone may cause breakdown of the skull base. Delay in the diagnosis of such injury may cause gradual herniation of the intracranial contents through the skull base defect and encephalocele formation and, life threatening complications including ascending meningitis, intracranial abscess and pneumocephalus. Therefore, surgeon should consider the possibility of such injury and seek for its findings.

Conflict of Interest: No conflicts declared.

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