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Meningoencephalocele Formation after Nasal Septoplasty and Management of this Complication

Nasal Septoplasti Sonrası Oluşan Meningoensefalosel ve bu Komplikasyonun Yönetimi

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ABSTRACT

OBJECTIVE: Patients undergoing nasal septoplasty can face many complications. Some of these complications are rare but their results are life threatening. Being aware of this complication could prevent further problems such as enlargement of the bone and dural defect, herniation of the meninges and brain tissue through the defect by pulsation of the brain and ascending infection. With early diagnosis, a less aggressive method could be used to treat this complication.

CASE DESCRIPTION: A 50-year-old woman was admitted our hospital with the complaint of loss of consciousness. Her Glasgow coma score was 7 on admission. She had no lateralizing signs, but had nuchal rigidity. Blood pressure was 200/110mm Hg, the respiratory pattern was apneic, complete blood count revealed 12000 leucocytes/mm3 and arterial blood gases showed respiratory acidosis and other biochemical parameters were within normal limits. Computerized cranial tomography (CCT) showed diffuse brain edema without evidence of other signs. Lumbar puncture was performed revealing purulent and highly viscous cerebrospinal fluid (CSF).

CONCLUSION: Late diagnosis and late repair of arachnoidodural tearing could lead to life-threatening complications, and cases with meningitis and larger defects may require more extensive surgery instead of transnasal endoscopic repair.

KEY WORDS: Meningoencephalocele, Meningitis, Nasal septoplasty, Rhinorrhea

ÖZ

Nasal septoplasti operasyonu yapılan hastalarda bazı komplikasyonlar gelişebilir. Bu komplikasyonlar nadir olmakla birlikte, bazen hayatı tehdit eden komplikasyonlarda ortaya çıkabilir. Septoplasti sırasında kripriform plate kırılması, dura defekti ve bazen araknoid defekti oluşması, oluşan defektten beyin ve meninkslerin herniasyonuna yol açar. Kemik defektin beyin pulsasyonu ile büyümesi, beyin ve zarlarının defektten herniye olması, asendan infeksiyon gelişmesi görülebilecek komplikasyonlardır. Bu komplikasyonların bilinmesi ve erken tanı konulması ile kısmen noninvaziv yollarla tedavi mümkündür. Vaka Sunumu: 50 yaşında kadın bilinç kaybı nedeni ile hastanemize kabul edildi. Kabul sırasında hastanın Glaskow Koma skoru 7/15 ve lateralize edici bulgusu yoktu, fakat ense sertliği mevcuttu. Kan basıncı:200/110 mm/Hg ve solunumu apneikti. Biyokimyasal ölçütleri normal sınırladaydı. Beyin tomografisinde yaygın ödem dışında bulgu yoktu. Lumbar fonksiyon yapılan hastanın beyin omurilik sıvısı pürülan görünümdeydi. Sonuç:Nasal septoplasti sonrası gelişebilecek komplikasyonların geç tanılanması hayatı tehdit eden komplikasyonlara yol açabilir. Menenjit gelişmiş ve geniş kemik defekti olan olgularda endoskopik transnasal onarıma ek olarak transkraniyal onarımda gerekli olabilir.

ANAHTAR SÖZCÜKLER: Nasal septoplasty, Meninjitis, Meningoensefalosel, Rhinorrhea

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INTRODUCTION

Complications such as septal abscess, septal perforation, hemorrhage, cavernous sinus thrombosis, meningitis, pneumatocephalus and rhinorrhea have been recorded during and after nasal septoplasty. Some of these complications are rare but their results are life threatening (8). The perpendicular crista galli it forms a relatively thick and strong bone complex when compared to the thin ethmoid labyrinth while the junction between the cribriform plate and the ethmoid labyrinth is particularly vulnerable to traumatic injuries as the bone here is, in contrast, quite delicate and the dura is tightly adherent (6,10,12). The cribriform plate supports the olfactory bulbs and is perforated by numerous foramina for the passage of the olfactory filaments. The closely adherent dura and small arachnoid pouches follow the olfactory filaments inside the ethmoid bone and fractures of the ethmoid bone are therefore likely to be associated with injuries to the arachnoid and olfactory filaments. (10,12). Exposure to the anterior skull base, especially around the cribriform plate through the sinonasal tract using the endoscopic method provides a noninvasive and effective way for repairing any defect around this zone. This method has been gaining in popularity among ENT surgeons and neurosurgeons in the recent years. In this report, a case of meningoencephalocele complicated by purulent meningitis is described and possible surgical treatment methods are reviewed based on the available literature and our experience from this case.

CASE REPORT

A 50-year- old female patient was admitted our hospital with the complaint of loss of consciousness. Her Glasgow coma score was 7 on admission. She had no lateralizing signs, but had nuchal rigidity. Blood pressure was 200/110mm Hg, the respiratory pattern was apneic, complete blood count revealed 12000 leucocytes/mm3 and arterial blood gases showed respiratory acidosis and other biochemical parameters were within normal limits. CCT showed diffuse brain edema without evidence of other signs. The patient was intubated and connected to the ventilator. Lumbar puncture was performed revealing purulent and highly viscous cerebrospinal fluid (CSF). Biochemical and microbiological analysis of CSF revealed bacterial meningitis and the bacterial CSF culture grew Streptococcus pneumonia. Antibiotic treatment was started and the patient's clinical condition improved within a week. She regained full consciousness and was extubated. Her medical history revealed that a nasal septoplasty operation had been performed ten years ago. She had experienced intermittent nasal and pharyngeal discharge since the nasal septoplasty operation. She had been offered transnasal endoscopic repair several times but had refused the operation. MR imaging of the cranium and paranasal sinus computerized tomography revealed a meningoencephalocele in the left nasal cavity (Figure 1-2). Over the next 20 days, antibiotic treatment was completed and the patient was discharged in normal neurological condition again after being offered surgery. The patient was operated three months later. Initially, a bifrontal craniotomy was performed and base of the anterior fossa and cribriform plate was reached, and there was 0.6x1.7 cm defect on the left side of the cribriform plate. The arachnoidal fold and a part of gyrus orbitalis were herniated through the defect into the ethmoidal air cell, and the sac formed from this herniation was sectioned from the entry point to the left cribriform plate. The ragged dura and the defect in the cribriform plate were repaired. In the second stage of the operation, the brain tissue obliterating the left nasal cavity was identified and removed endoscopically through the transnasal route by the ENT colleagues. Macroscopically, the meningoencephalocele was a pale-colored sac about

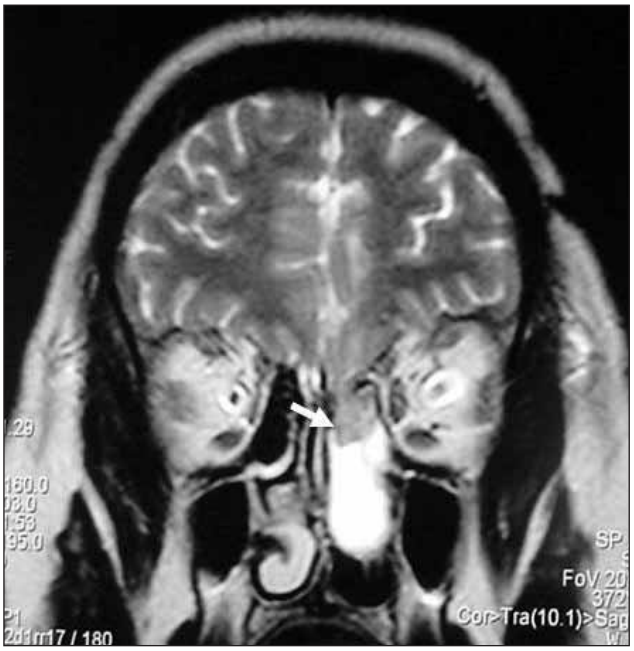

Figure 1: *T2-Weighted MR image. Arrow indicates partly herniated brain and CSF filled sac through the defect.*

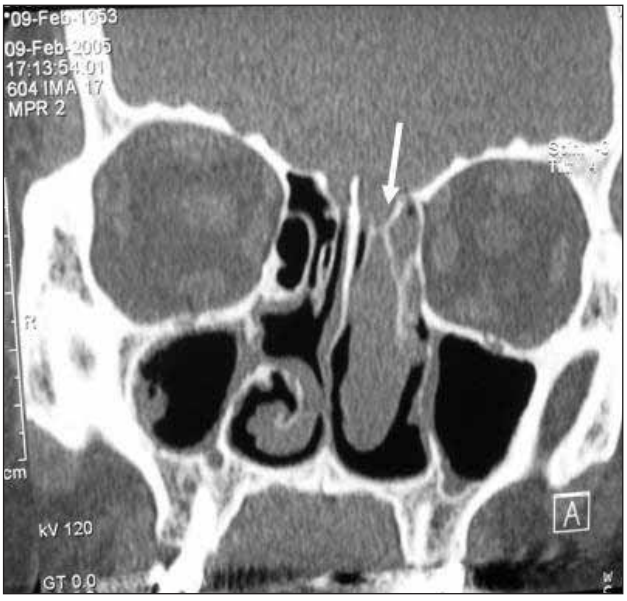

Figure 2: *Paranasal sinus computed tomography. Arrow indicates the defect in the left cribriform plate and the lesion.*

3x1 cm in size and was sent for pathologic analysis. The defect was visualized from the cranial and nasal cavities. The defect of the cribriform plate was repaired by free bone graft prepared from the middle turbinate and the galea graft settled intracranially on the visualized defect. In addition, a mucosal flap prepared from the middle turbinate was rotated endoscopically to repair the cribriform defect intranasally. The biopsy result showed neurogliotic tissue (Figure 3) and abscess formation (Figure 4). The patient was put on bed rest and diuretics. She did not have any rhinorrhea after the operation. She was discharged with full recovery.

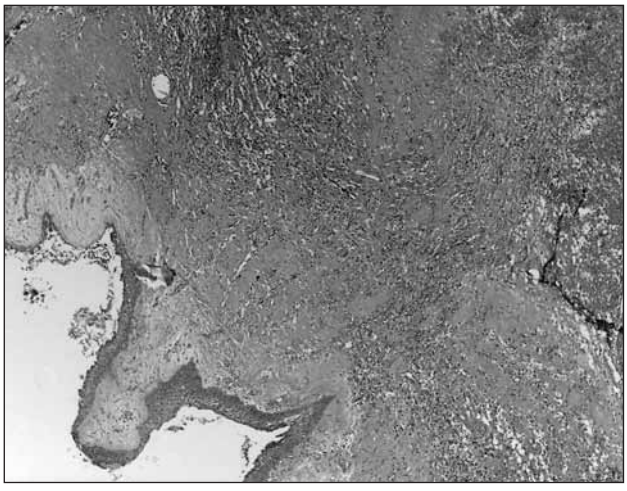

Figure 3: *Neuroglial cells and pseudostratified respiratory epithelium (HEx100).*

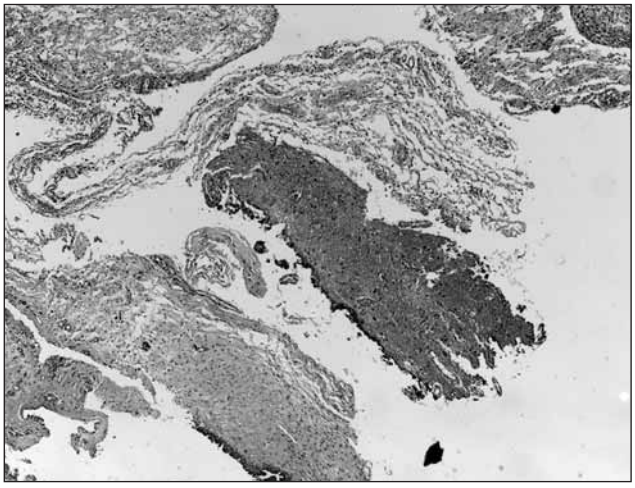

Figure 4: *Pseudostratified respiratory epithelium with extensive squamous metaplasia and foci of abcess (HEx40).*

The patient was periodically controlled endoscopically and showed no evidence of recurrence and rhinorrhea on her last follow-up 38 months after surgery.

DISCUSSION

Rhinorrhea is a discharge of cerebrospinal fluid due to a defect in duramater and a bone defect in the base of the anterior part of the skull base. Pathological connections appear mainly at the weaker bone structure (the roof of the frontal, ethmoid, sphenoid sinus and cribriform plate). An anterior skull base defect would cause an ascending meningitis. (7,8). Cakmak and associates defined the technical difficulties and prevention of the rhinorrhea after endoscopically-assisted transsphenoidal pituitary surgery, but they did not mention any cribriform plate injury during transsphenoidal surgery (2). Taveras and Ransohoff proposed the following mechanism for the formation of a LMC: trauma produces a dural tear and skull fracture; the leptomeninges then herniate through the dural defect; the herniating leptomeninges preclude normal healing and apposition of the bone defect at the base of skull. Pulsation of the brain gradually pushes more leptomeninges through the defective part of the dura; subarachnoid fluid becomes trapped in the herniated tissue probably secondary to arachnoidal adhesions and forms a cyst, and the growing cyst then gradually erodes the edges of the bone and compresses the underlying cortical bone leading to atrophy. Dura mater provides nutritional support to the skull, so any deficit of duramater disturbs the blood supply of the

bone and bone healing and osteoblastic activity (11). We think that the theory of the Taveras and Ransohoff explains the acquired meningoencephalocele after nasal septoplasty operation taking into account arachnoidal tearing together with dura mater tearing during nasal septoplasty procedure. There are two possible mechanisms for the occurrence of CSF fistula after nasal septoplasty operation. The first is that the elevator forceps may be moved too superiorly beyond the limits of ethmoid roof during elevation of septal mucoperichondrium at the superior tunnel. The second possible mechanism is the creation of fractures in the cribriform plate by fracturing the perpendicular lamina (8). In view of neurosurgery, there may be a different benign lesion involved in the nasal cavity such as nasal glial heterotopia, encephalocele, meningocele and meningoencephalocele. Nasal glial heterotopia is a mass composed of mature brain tissue isolated from the cranial cavity or spinal canal. Nasal glial heterotopia is frequently diagnosed in newborn infants; however, it may rarely be found in adults. These benign congenital tumors are usually found in the nasal region (1,3,9), and macroscopically nasal glial heterotopia has no connection with the subarachnoid space and brain, but encephalocele is a part of brain tissue that herniates through an osseous defect due to the pulsatile effect of the brain, and is connected to the rest of the brain by a pedicle. Nasal glial heterotopia could not cause rhinorrhea, but encephalocele, meningocele and meningoencephalocele may cause meningitis and rhinorrhea. The cranial MRI and parasellar computerized tomography of the patient demonstrated connection between the intranasal lesion and brain. This connection provided to differentiate between NGH and the lesions connected to brain such as meningoencephalocele and leptomeningeal cyst both could cause rhinorrhea and meningitis but not nasal glial heterotopia. Histologically, nasal glial heterotopia and meningoencephalocele contain neural and glial tissue but leptomeningeal cyst does not contain neural tissue (4,5,7,8). We suppose that our presented case could be accepted as an acquired meningoencephalocele rather than nasal glial heterotopia or leptomeningeal cyst regarding pathology specimen, patient medical history, cranial CT and cranial MRI. In this case, we preferred to perform combined surgery (transnasal and transcranial) in contrast to the recent trends among ENT surgeons and neurosurgeons. Wigand first introduced endoscopic closure of the CSF leaks through the transnasal route in 1981. After introduction of these techniques, ENT surgeons and neurosurgeons prefer to repair CSF leaks using this technique with a high success rate. Hydrocephalus and meningitis are the common causes of failure for transnasal endoscopic repair of CSF leaks. In this report, a case of iatrogenic meningoencephalocele complicated by purulent meningitis that was causing a life-threatening clinical condition, could pose more risk than any simple iatrogenic fracture of the cribriform plate that could easily be repaired through the sinonasal route using an endoscopic technique. Zweig et al. reported 17 cases with encephalocele, meningoceles and meningoencephaloceles treated through the transnasal route using an endoscopic technique. According to their results, the location and size of the skull base defect, its etiology, and the technique and choice of material used for repair did not significantly affect the surgical outcome. However, the presence of hydrocephalus was significantly related to poor surgical outcomes. There were only two patients without a satisfactory result in their series: one suffered from repeated meningitis with posttraumatic hydrocephalus and the other one also had hydrocephalus without meningitis. Intracranial approaches have some potential risks such as anosmia, frontal lobe retraction, and intracranial hemorrhage. Furthermore, failure rates for CSF rhinorrhea can be as high as 30%. Although transnasal endoscopic repair is the current treatment method for repairing CSF fistulas, the combined approach (transnasal endoscopic repair and transcranial repair of the defect) may be more appropriate in cases with rhinorrhea presenting with meningitis and an obvious anterior cranial base defect. In addition, the intracranial approach allows full exposure of the anterior fossa and satisfactory exploration of the cribriform plate, sphenoid wing and orbital roof. It is possible to resect the herniated brain, dissect the dural margins and repair the defective bone edges under direct vision. Extracranial approaches may not be successful in the presence of gliotic brain and ragged dural margins (5). We preferred combined surgery because of two reasons; prevention of ascending meningitis due to postoperative leakage, which is one of the important

consequences of the failure in transnasal endoscopic repair of the roof of the ethmoid bone, and complete resection of degenerating neurogliotic tissue and ragged dura mater. The pathology specimen also showed neurogliotic tissue and abscess formation (Figure 3-4). The patient has been followed-up periodically. Her last examination showed no rhinorrhea or recurrence.

CONCLUSIONS

Traumatic basal skull fracture and iatrogenic injury during nasal septoplasty are the main causes of cerebrospinal fluid fistulas in adults. The roof of the ethmoid bone and the cribriform plate are the most frequent sites of rhinorrhea. The technique for repairing CSF leaks has evolved from intracranial repair to extracranial approaches. The transcranial approach could be added to the endonasal technique to prevent recurrent meningitis in patients with large defects as in the presented case.

REFERENCES

1. Chan JK, Lau WH: Nasal astrocytoma or nasal glial heterotopia? Arch Pathol Lab Med 113: 943-945, 1989
2. Çakmak Ö, Bavbek M, Caner H, Akkuzu B, Ergin T, Altınörs N: Endoskop yardımlı transsfenoidal hipofiz cerrahisinde teknik güçlükler ve dikkat edilmesi gereken noktalar. Türk Nöroşirürji Dergisi 13: 40-46, 2003
3. Ersahin Y, Gurmen V, Palali I: Growing skull fractures (craniocerebral erosion) S. Neurosurg Rev. 23: 139-144, 2000
4. Gupta V , Sinha S, Singh AK, Kumar S, Singh D: Growing skull fracture of ethmoid: A report of two cases J Maxillofac Surg.28: 224-228, 2000
5. Lewin W: Cerebrospinal fluid rhinorrhoea in non missile head injuries Clinical Neurosurg 12: 237–252, 1966
6. Muhonen Michael G, Piper John G, Menezes Arnold H: Pathogenesis and Treatment of Growing skull Fractures. Surg Neurol 43: 367-373, 1995
7. Onerci TM, Ayhan K, Ogretmenoglu O: Two consecutive cases of cerebrospinal fluid rhinorrhea after septoplasty operation. Am J Otolaryngol 25:354-356, 2004
8. Park J I, Strelzow V V, Friedman W H: Current management of cerebrospinal fluid rhinorrhoea. Laryngoscope 93: 1294–1300, 1983
9. Penner CR, Thompson L: Nasal glial heterotopia: A clinicopathologic and immunophenotypic analysis of 10 cases with a review of the literature. Ann Diagn Pathol 7: 354-359, 2003
10. Taveras J M, Ransohoff J: Leptomeningeal cysts of the brain following trauma with erosion of the skull. A study of seven cases treated by surgery. J Neurosurg 10: 233–241, 1953
11. Wigand ME: Transnasal ethmoidectomy under endoscopical control. Rhinology Mar 19(1): 7-15, 1981
12. Zweig JL, Carrau RL, Celin SE, Snyderman CH, Kassam A, Hegazy H: Endoscopic repair of acquired encephaloceles, meningoceles, and meningo-encephaloceles: predictors of success. Skull Base 12: 133-139, 2002