Case Report

Spontaneous Rhinorrhea Associated with Defect at Cribriform Plate and Orbital Roof Seen with Primary Empty Sella Syndrome: Case Report

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Summary

Rhinorrhea which is usually seen after trauma and surgical interventions but rarely seen spontaneously may lead to mortal complications if not treated. Traumatic rhinorrhea usually ceases without surgery however since this is not true for non-traumatic rhinorrhea, it requires surgical intervention. Spontaneous rhinorrhea is generally associated with empty sella and in these cases osseous defects are detected frequently in the walls of the paranasal sinuses and skull base. Although in these cases signs of raised intracranial pressure such as empty sella are usually seen, rhinorrhea can also be seen without raised intracranial pressure. Here we report a 46-year old woman presented complaining of clear fluid leaking from her nose who had defect of cribriform plate and orbital roof together with empty sella and was operated with the diagnosis of spontaneous rhinorrhea. In course of postoperative follow-up for eighteen months no rhinorrhea was seen.

Key words: Spontaneous rhinorrhea, Cribriform plate, Orbital roof, Primary empty sella syndrome

Özet


Anahtar Kelimeler: Spontan rinore, Kribriform plate, Orbita tavani, Primer empty sella sendromu
INTRODUCTION
Rhinorrhea is a rare disease generally seen after trauma and surgical interventions. However, non-traumatic rhinorrhea, in another word ‘spontaneous rhinorrhea’ is more rare and constitutes 4-20% of all rhinorrhea cases (5,9,15).

The first non-traumatic rhinorrhea case was reported by Miller in 1826, St.Clair Thomson reported review of spontaneous rhinorrhea cases in 1899 followed by Aubin et al. reporting a case with a small defect in lamina cribrosa in 1944 (9). Cases and reviews continued to be reported thereafter.

Rhinorrhea should be treated surgically since it may lead to mortal complications such as meningitis, intracranial abscess and spontaneous pneumocephalus if not treated (3,13).

The relationship between empty sella and spontaneous rhinorrhea was first described by Ommaya et al. (9). The rhinorrhea associated with primary empty sella syndrome is generally together with fistulae at sellar region (7,10,12).

However rhinorrhea cases which are together with primary empty sella syndrome and osseous defects of lamina cribrosa and orbital roof are very rare (10,14). We report a spontaneous rhinorrhea case associated with primary empty sella syndrome together with osseous defect of lamina cribrosa and orbital roof.

CASE PRESENTATION
46-year-old female patient presented with two-month history of clear fluid leaking from the right nostril and headache who had been treated for allergic rhinitis and sinusitis. The leakage was increasing while bending forward. She had no history of cranial trauma and surgery. Neurological examination was normal.

Nasal fluid contained 61 mg/dL of glucose (≥30 mg/dL consistent with CSF) and serum glucose level was 88 mg/dL (normal limits:70-110 mg/dL) and the results were consistent with CSF. Serum hormones (T3, T4, TSH, FSH, LH, prolactin, ACTH, cortisol, GH) were within normal limits.

Empty sella was detected in cranial and sellar MRI scans (Figure 1). Ventricule sizes were within normal ranges. CSF fistulae at cribriform plate and orbital roof were documented in dynamic MRI (Figure 2). Osseous defects were detected in cribriform plate and orbital roof in cranial CT scan (Figure 3).

Bifrontal craniotomy procedure was performed using intradural approach. During the surgery, herniations of dura together with brain tissue through the osseous defects at cribriform plate and orbital roof were seen (Figure 4). The herniated dura and brain tissue were excised and dura was repaired. Osseous defects at cribriform plate and orbital roof were repaired using fibrin glue, autogenous bone and muscle grafts. No intervention was performed for empty sella.

The lumbar spinal drainage catheter inserted peroperatively was taken out in the end of the fifth day and the patient was discharged with no neurological deficit on the seventh day of postoperative period. In course of follow-up for eighteen months no rhinorrhea or any other complaint was seen.
**Figure 1:** T1 and T2-weighted MR images of brain show empty sella.

**Figure 2:** Dynamic MR images show CSF leakage and brain tissue herniating through defects of cribriform plate and orbital roof.
DISCUSSION

Rhinorrhea is divided into two as traumatic and non-traumatic. Traumatic rhinorrhea is subdivided into two as accidental and iatrogenic while non-traumatic rhinorrhea is subdivided as normal pressure and high pressure leaks\(^{(9)}\). \%4-20 of rhinorrhea cases are non traumatic\(^{(5,9,15)}\). Traumatic rhinorrhea usually ceases spontaneously. However spontaneous rhinorrhea is treated surgically because of presence of osseous defects at skull base and paranasal sinuses which is usually together with empty sella\(^{(4,6,10)}\).

In spontaneous rhinorrhea cases; osseous defects are localized at lateral sphenoidal...
recess, central sphenoidal recess, ethmoidal roof, lamina cribrosa, frontal sinus, posterior frontal recess and supraorbital ethmoidal bone. In our case different from the literature, the osseous defects are at lamina cribrosa and orbital roof together.

Schlosser et al. documented empty sella in 100% of spontaneous rhinorrhea cases. They advocated that rhinorrhea occurs secondary to primary empty sella syndrome with the theory that CSF pulsations caused by raised intracranial pressure compress the hypophysis against the floor of sella and this leads to CSF fistula by eroding the diaphragma sella and the bone. In our case sellar floor was intact in spite of the presence of empty sella.

In impaired CSF absorption theory Brismar and Bergstrand emphasized that the obstruction of CSF circulation at the level of the arachnoid villi increases intracranial pressure and powerful CSF pulsations erode sellar floor and diaphragma sella and this causes CSF leakage through the osseous defects. In focal atrophy theory; atrophy of cribriform plate, olfactory nerve and sellar structures causes the dura together with brain tissue herniating through the ruptured arachnoidal projections. We did not see olfactory nerve atrophy peroperatively.

Shetty et al. emphasized that extensive pneumotization of the paranasal sinuses diminishes the thickness of the bony walls and raised intracranial pressure forces CSF to leak through the osseous defects at the thinnest site of the bone. Increased pneumotization was detected at the lateral recess of the sphenoidal sinus in 91% of the spontaneous rhinorrhea cases. The fact that lateral recess is not affected in any case of traumatic rhinorrhea despite the most frequent localisation of CSF fistula being at the lateral recess in non-traumatic rhinorrhea supports this theory. Intracranial pulsatile pressure causes arachnoidal herniations through these thinned walls, creating encephaloceles thereafter and leading to rhinorrhea. In our case we documented the herniation of the dura and brain tissue through the osseous defects at lamina cribrosa and orbital roof supporting this theory.

Herniation of the dura and brain tissue through the congenital osseous defects at sellar floor secondary to CSF pulsations is another factor causing spontaneous rhinorrhea.

Rhinorrhea was also reported during the medical treatment of the prolactinoma. In the prolactinoma cases treated with dopamine agonists, rhinorrhea occurs because of reduction in the tumour size usually after two years of treatment. There is no history of use of dopamine agonists in our case.

Schlosser et al. identified 81% of the spontaneous rhinorrhea cases as obese, middle-aged females and mean body mass index was 37.7. Our case was a middle-aged female and her body mass index was 37 similar to literature.

It is advocated that central obesity increases intraabdominal pressure and indirectly intracranial pressure causing primary empty sella syndrome. However Brismar and Bergstrand opposed this argument because of the rarity of the spontaneous rhinorrhea in spite of the high frequency of the central obesity in the society. Nevertheless since rhinorrhea can also be seen secondary to congenital osseous defects at skull base the contribution of the central obesity to rhinorrhea could be accepted.

Despite the fact that CT is superior to MRI to detect the osseous defects, coronal and sagittal MRI shows the exact sites of osseous defects and even tiny CSF leaks successfully because of the contrast signal intensities of the bone and the CSF. In our case, CT and MRI depicted defects at lamina cribrosa and orbital roof together with soft tissue herniations through these
defects which were also documented peroperatively.

In conclusion; we reported a case of spontaneous rhinorrhea associated with defects at cribriform plate and orbital roof together with primary empty sella syndrome which is very rare in the literature. We theorize that this association is a result of constant CSF pulsations leading to primary empty sella syndrome and herniation of dura and brain tissue through osseous defects.

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Received by: 26 January 2012
Revised by: 13 May 2012
Accepted: 14 May 2012

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