Non-traumatic High-Pressure Cerebrospinal Fluid Rhinorrhea as an Initial Presentation of Tentorial Base Meningioma- Case Report and Review of the Literature

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Abstract:

**Background:** Patients with posterior fossa tumor usually presents with headache, vomiting, papilledema along with other features of focal neurological deficit. The mass effect is evidenced due to development of hydrocephalus as a consequence of blockage of cerebral aqueduct. However, as a sequence of long standing hydrocephalus, there is possibility of erosion of cribriform plate which may give rise to CSF rhinorrhea. Though spontaneous CSF rhinorrhea is a common presentation of the tumor of anterior cranial base but it’s a very rare manifestation of posterior fossa tumor as an initial presentation.

**Aim of the work:** The aim of this case report is to describe a very rare case of tentorial base meningioma which initially presents as spontaneous CSF rhinorrhea. The possible cause of the rhinorrhea and the relationship with the tumor are analyzed.

**Case Report:** A 35-year-old normotensive, non-diabetic female presented with spontaneous watery nasal discharge through her left nostril following which she developed giddiness along with scanning speech. After thorough radiological evaluation, she was diagnosed as a case of left sided tentorial base meningioma with erosion of cribiform plate of ethmoid bone. At first sitting, she underwent left sided suboccipital craniotomy and gross total removal of tumor. 2 months later, she underwent bifrontal craniotomy, repair of dural defect and reconstruction of bone gap with titanium mesh. Her post-operative period was uneventful. She was discharged to home without having any neurological deficit.

**Conclusion:** CSF rhinorrhea is a rare manifestation of chronically raised intracranial pressure secondary to posterior fossa tumor.

**Key words:** non-traumatic CSF rhinorrhea, tentorial base meningioma.


Introduction:

Cerebrospinal fluid (CSF) rhinorrhea commonly occurs after head injury, usually as a sign of fracture of anterior cranial base. It may also find after removal of sellar and suprasellar tumor through trans-sphenoidal approach. Spontaneous CSF rhinorrhea is rarely found in brain tumor, specially before surgical excision. CSF leaks have been reported to be associated with meningiomas of anterior cranial base, craniopharyngiomas, invasive pituitary adenomas, osteomas and nasopharyngeal tumors. The rhinorrhea occurs as a result of direct neoplastic infiltration and destruction of the dura and intervening bone between subarachnoid space and the nasal cavity. CSF rhinorrhea caused by infratentorial tumors are rarely reported in the literature. Ommaya has described this type of CSF leak as an “indirect” type of “non-traumatic” fistula. The fistula results from increased intracranial pressure due to long standing hydrocephalus -causing erosion of the basal dura.

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along with anatomically fragile areas of the skull base. The cribriform plate area and the sellar floor are the most vulnerable areas in this aspect. Our literature search revealed a few case reports of such non-traumatic spontaneous CSF fistulae as a consequence of raised intracranial pressure due to intracranial tumors. Here, we report a rare case of non-traumatic CSF rhinorrhea which was the initial presentation of a large tentorial base meningioma.

**Case Report:**

*History and physical examination*

A 35 years old normotensive, non-diabetic lady presented with watery nasal discharge for 1-year, occasional headache for 6 months, progressive dimness of vision and ataxia for 2 months. After neurological examination, her visual acuity was 6/6 in both eyes and fundoscopy revealed papilledema. Patient had truncal ataxia with tendency to fall towards left. Rest of the cranial nerves were intact. Motor and sensory system revealed no abnormalities. After admission, her nasal discharge stopped spontaneously. However, after excision of tumor rhinorrhea reappeared. By this time, we clinically confirmed the discharge as CSF and she diagnosed as a case of non-traumatic CSF rhinorrhea.

*Investigations*

After thorough radiological evaluation, patient diagnosed as a case of left sided tentorial base meningioma. A fairly large T1WI isointense and T2WI hyperintense extra axial tumor, measuring about 6 * 5.5 * 5 cm attached with tentorium cerebelli, having significant mass effect evidenced by compressing the tentorial surface of left cerebellar hemisphere with obstruction of cerebral aqueduct, giving rise to triventricular hydrocephalus. After giving contrast, the tumor had homogenous contrast enhancement with dural tail along the tentorium (figure 1A, B).

**Operative procedure**

With all aseptic precaution, patient underwent left suboccipital craniotomy and gross total removal of tumor (simpson grade 1). A hockey stick incision was given over left occipital region. After subperiosteal dissection, a 4x4 cm craniotomy done. Dura incised in C shaped fashion, base directed towards transverse sinus. Tumor was greyish red in color, soft, suckable with high vascularity. Gross total removal done following four principle of meningioma surgery. Hemostasis ensured and closure done in standard fashion.

**Post-operative period**

Patient’s recovery was uneventful. Resected specimen was sent for histopathological examination and biopsy showed transitional meningioma (WHO grade I).

**Hospital course**

15 days after the operation, she developed watery nasal discharge which was salty, crystal clear, doesn’t produce any stain on tissue paper, aggravated during sitting, bending forward, coughing and relieved after lying down. We measured sugar level of the discharge as a bedside test of CSF rhinorrhea. After that, CT cisternography done which showed a 12.2 * 9.1 mm bone gap through left sided cribriform plate of ethmoid bone (figure 2A, B). She diagnosed as a case of non-traumatic CSF rhinorrhea.

![Fig.-1: MRI of brain T1WI sagittal section showing a massive tentorial base meningioma with gross dilatation of the lateral ventricle(A), After administration of contrast, there is homogeneous contrast enhancement (B).](image)

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non-traumatic CSF rhinorrhea. Again, she underwent bifrontal craniotomy and repair of the site of leakage by dural closure, fat graft, augmented by tissue glue and previously harvested pericranium as overlay. Bone gap was reconstructed by titanium plate (figure 3A-D). Post-operative CT scan with bone window showing no tumor residual (figure 4A) and proper position of the titanium plate (figure 4B).

Fig.-2A, B: CT scan of brain with bone window showing a 12.2 *9.1 mm bone gap in the cribriform plate of ethmoid bone (left side).

Fig.-3: Per-operative photographs showing the bone defect in the cribriform plate (A), watertight dural closure with anterior cranial base reconstruction with fat (B), harvested pericranium as overlay (C), bone flap fixed with miniplate and screws(D).
Discussion:
The term “spontaneous” cerebrospinal fluid fistula was first described by Miller in 1826 [1], which later on replaced by Ommaya in 1968 who suggested that “non-traumatic” cerebrospinal fluid fistula would be more accurate term and classified this entity into high pressure and normal pressure categories [2]. Non-traumatic high-pressure CSF fistula is uncommon. It can result from direct erosion of anterior cranial base by the tumor itself or indirectly as a consequence of raised intracranial pressure due to hydrocephalus resulting from blockage of CSF pathway by large tumor of remote origin. The cribiform plate and basal dura of the anterior cranial base are the most vulnerable area to be affected. Developmental and anatomical factors may predispose to this increased vulnerability for some individuals. CSF rhinorrhea, in such cases, may occur both before and after tumor excision and classified into pretreatment and post-treatment types [3].

In 1978, Oblu [4] identified several causes of non-traumatic spontaneous cerebrospinal fluid leak and classified as follows:

- congenital: congenital defects of anterior skull base especially to laminar cribriforma with or without meningoencephaloceles, cranio stenosis (Crouzon disease, oxycephaly), Arnold Chiari syndrome, hydrocephalus by congenital stenosis of aqueduct of Sylvius.

- tumoral: by direct mechanism (bonedefect generated by compression or infiltration) or indirect mechanism (intracranial hypertension as a consequence of hydrocephalus)

- infectious: parasitic diseases, tuberculous meningitis, osteomyelitis

- vascular: ethmoidal vessels thrombosis

- empty sella syndrome

Non-traumatic CSF rhinorrhea accounts for less than 5% among all types of CSF fistula [4]. Among them infratentorial tumors remote from the site of CSF fistula were infrequently reported in the previous literature. Jing-ju et al reported a tentorial base meningioma causing CSF rhinorrhea in 2007 [5]. Other reported infratentorial tumors are choroid plexus papilloma of the fourth ventricle [6] and fourth ventricle ependymoma [7]. Topographically posterior fossa meningioma consists of cerebellar convexty, cerebello-pontine angle, tentorial, jugular foramen, petroclival, foramen magnum, IV ventricle and unclassified. Commonly these tumors presented with the features of ataxia, hydrocephalus, signs of cerebellar dysfunction or multiple cranial nerve palsies. In our reported case, initial presentation was CSF rhinorrhea which was mistaken as common cold- for which patient did not seek any medical attention. 5 months later, when she developed headache and ataxia, she diagnosed as a case of tentorial base meningioma after thorough radiological evaluation. However, after excision of tumor- rhinorrhea subsided for a short period of time. When she again developed similar symptom, we clinically confirmed the nasal secretion as CSF by measuring its sugar level.

![CT scan of brain showing evidence of previous suboccipital craniotomy with gross total removal of the tumor (A), Bone window reveals reconstruction of the cribiform plate with titanium mesh (B).](image-url)
of leakage was identified by CT cisternogram following which she underwent definitive surgery.

Regarding pathogenesis non-traumatic CSF rhinorrhea, the aetiology is either congenital or acquired. There are several embryological variations of cribriform plate and the traversing olfactory fibres for which minor trauma or forceful sneezing may result in CSF rhinorrhea. The CSF fistula in the reported case was probably related to the presence of the large intracranial tumor causing obstructive hydrocephalus resulting in chronically raised intracranial pressure. After certain period, this raised intracranial pressure resulted in plugging of the frontal lobe into the site of the fistula which results in spontaneous cessation of the rhinorrhea. After excision of tumor, the intracranial pressure become normalizes for which rhinorrhea reappears after first operation. So, our case possesses both pretreatment and post treatment component according to Liang's classification. In management of such cases, Liang recommended that patients of the pretreatment type should be managed conservatively or with CSF diversion procedure, while patients of the post-treatment type need direct repair of the fistula. For this reason, our patient underwent bifrontal craniotomy and repair of the fistula in the second sitting.

Conclusion:
CSF rhinorrhea is very rare presentation of infratentorial tumor as an initial presentation without having any sign of focal neurological deficit.

References: