

Spontaneous Cerebrospinal Fluid Rhinorrhea Indirectly Caused by Brain Tumor

—Report of Two Cases and Review of the Literature—

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Abstract

Two cases of nontraumatic cerebrospinal fluid rhinorrhea were presumed to be an indirect manifestation of brain tumor. One patient had a parietal convexity meningioma and the other a cerebello-pontine angle neurinoma. Following excision of the tumors these patients spontaneously developed cerebrospinal fluid rhinorrhea, which disappeared after the repair of fistulas at the base of the frontal fossa. In such cases the rhinorrhea may be caused by fragility of certain structures as a consequence of long-standing increased intracranial pressure due to brain tumor.

Key words: cerebrospinal fluid rhinorrhea, intracranial fistula, convexity meningioma, cerebellopontine angle neurinoma, cribriform plate

Introduction

Brain tumors that directly invade the nasal cavity sometimes cause cerebrospinal fluid (CSF) rhinorrhea.¹⁾ However, CSF rhinorrhea as a manifestation of a remote brain tumor has rarely been documented in the literature.^{2,4-15,17)} We encountered two instances of CSF rhinorrhea *via* the base of the frontal fossa, each of which occurred after excision of a tumor that had not invaded the nasal cavity. One was a parietal convexity meningioma and the other was a cerebellopontine angle neurinoma. The literature concerning CSF rhinorrhea as an indirect manifestation of brain tumor is reviewed and its pathogenesis is discussed.

Case Reports

Case 1: A 49-year-old female was admitted to our hospital on July 29, 1971, following the sudden onset

of Jacksonian epilepsy. The seizure started at the left upper extremity and eventually spread to the entire body. After the first convulsion, she noted weakness of the left upper and lower extremities. She had several seizures on the day of admission.

The general physical examination was normal except for slight kyphosis of the thoracic spine. The neurological examination revealed slight left hemiparesis without spasticity and diminished sensation in the left lower extremity. A fundoscopic examination disclosed advanced bilateral papilledema. Plain skull films showed erosion of the sellar floor, a sign of long-standing elevated intracranial pressure (Fig. 1A), and hyperostosis of the right parietal region. Carotid angiograms indicated a sunburst configuration in the right parietal convexity; it was supplied by the external and internal carotid arteries and corresponded to the hyperostotic area (Fig. 1B). A brain scan revealed a positive isotope accumulation in the same area.

A right parietal craniotomy was performed on August 17, 1971. Marked bleeding occurred while making the free bone flap, which encompassed the

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Fig. 1 A: Plain skull roentgenogram of Case 1 showing erosion of the sellar floor, possibly attributable to long-standing increased intracranial pressure. B: The lateral view of the right carotid angiogram disclosing the sunburst appearance of the parietal convexity meningioma, which is supplied by the enlarged, tortuous superficial temporal artery as well as the middle meningeal and anterior and middle cerebral arteries.

hyperostotic area. The tumor was round, soft, elastic, and bloody and measured approximately 5 cm in diameter. It was fairly well demarcated, from the surrounding edematous brain. Some portions of the mass adhered to the dura, and it was removed *en bloc* with the dura. The histological diagnosis was meningotheliomatous meningioma with occasional whorls and psammoma bodies.

During the uneventful postoperative course the left hemiparesis and papilledema gradually subsided. However, one month after surgery she noticed a watery rhinorrhea, which spontaneously disappeared after a few days. During 1975 she developed sporadic nasal discharge that spontaneously appeared and disappeared, but refused medical examination. She was readmitted on July 15, 1979, when she developed headache, fever, and confusion, on suspicion of meningitis caused by CSF rhinorrhea.

On her second admission she was febrile and confused with nuchal stiffness and Kernig's sign. However, the remaining neurological examinations were normal. The CSF obtained by lumbar puncture was turbid, with a cell count of $8960/\text{mm}^3$ (polymorphonuclear leukocytes, 86%; lymphocytes, 14%). The CSF contained 1340 mg/dl of protein and showed a strong, positive globulin reaction. The glucose was 17 mg/dl. The CSF culture yielded *α -Streptococcus*. Aminobenzyl penicillin was administered at 16 gm daily for 2 weeks and then at 8 gm daily for one additional week, after which the CSF cell count returned to normal. Plain skull films and tomograms disclosed no fistulas at the base of the skull. Radioisotope cisternography also failed to reveal the culpable fistula, but large amounts of radioisotope



Fig. 2 Intraoperative photograph of Case 1 showing a fistula at the right frontal base (arrowhead) and another at the left cribriform plate (arrow).

were collected on a cotton pledget in the left nasal cavity. After her complete recovery from the meningitis she was prepared for surgical repair of the CSF fistula. Throughout her second admission she was afflicted with a perplexing, severe cough.

A bifrontal craniotomy was performed on September 20, 1979. All procedures were performed *via* the intradural route. One fistula was found in the left cribriform plate and another at the right orbital roof, 1.5 cm to the right of the midline (Fig. 2). Each was about 2 mm in diameter. Portions of brain tissue had herniated into both fistulas. A patch consisting of falx plus embedding muscle was used to close the left fistula. The fistula of the right orbital roof was closed with a small piece of fascia lata. Finally, the entire frontal base was covered with a large piece of fascia lata.

The patient recovered uneventfully and had no further CSF leakage. Her severe cough subsided immediately after surgery. Six years after the second operation, the only sequela is anosmia.

Case 2: A 37-year-old male with a right-side hearing disturbance of nearly 12 years' duration experienced sudden onset of right tinnitus on January 4, 1978. Eighteen months later, following an otological examination, he was referred to our department for surgical management of an intracranial tumor.

The general physical examination was normal. The neurological examination revealed marked hearing disturbance and slight peripheral facial paresis on the right side. In the neuro-otological examination the caloric test disclosed palsy of the right canal. The routine laboratory investigation was normal. The CSF contained a high protein level (204 mg/dl) but a normal cell count ($1/\text{mm}^3$). The intracranial pressure was normal, with an opening pressure of 140

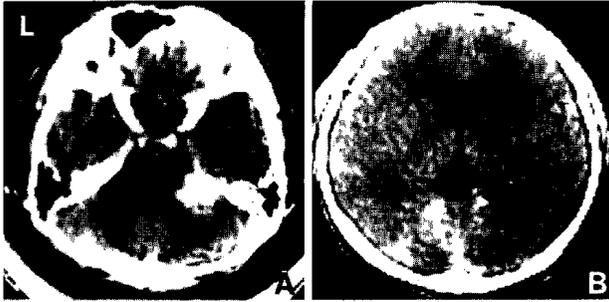


Fig. 3 A: Computed tomography after contrast enhancement in Case 2 demonstrating a high-density mass in the right cerebellopontine angle. B: Undiscernible cerebral sulci and slightly dilated lateral ventricles may indicate increased intracranial pressure.

mmH₂O. Plain skull films and tomograms disclosed erosion and widening of the right internal auditory meatus. Computed tomography revealed a mass at the right cerebellopontine angle, which was markedly enhanced by contrast media (Fig. 3A). The cerebral sulci were not visible and the lateral and third ventricles were slightly dilated, presumably due to increased intracranial pressure (Fig. 3B). Vertebral angiography indicated medial deviation of the right anterior inferior cerebellar artery, without tumor staining.

A right suboccipital craniectomy was performed on July 24, 1979. Exposure of the cerebellopontine angle revealed a soft, yellowish mass, from which a yellowish fluid was aspirated through a needle. The tumor was totally removed piece by piece under an operating microscope.

On the third postoperative day, the right facial paresis worsened slightly. A watery rhinorrhea was noted on the fourth postoperative day and he complained of frequent coughing. Radioisotope cisternography revealed abnormal accumulation of isotope in the right nasal cavity, although neither tomograms of the base of the skull nor computed tomography disclosed fistulas.

The patient underwent a right frontotemporal craniotomy on August 30, 1979. Intradural observation of the frontal base revealed a fistula measuring about 2 mm in diameter in the right cribriform plate (Fig. 4). The fistula was filled with Aron- α and then closed with the dural flap. The site of the previous suboccipital craniotomy was reopened in a search for additional fistulas. However, none were found in the right posterior cranial fossa or internal auditory meatus.

The CSF rhinorrhea ceased immediately after the



Fig. 4 An intraoperative photograph of Case 2 reveals a fistula at the right cribriform plate (arrow). The right olfactory nerve is also shown (arrowhead).

second operation and the cough disappeared. Six years postoperatively he remains well except for right hearing loss and slight facial paresis.

Discussion

Ommaya *et al.*⁸⁾ pathophysiologically classified non-traumatic (spontaneous) CSF rhinorrhea into that associated with high CSF pressure and that occurring with normal CSF pressure. In the presence of elevated pressure, CSF rhinorrhea is most often caused by tumor or hydrocephalus. Patients in whom an intracranial tumor has destroyed the base of the skull contiguous to the nasal cavity (such as occurs with pituitary adenoma and olfactory groove meningioma) occasionally develop CSF rhinorrhea before and/or after surgical removal of the tumor.¹⁾ On the other hand, intracranial tumors remote from the frontal base rarely cause CSF rhinorrhea,⁸⁾ the incidence being about 1% of all cases of CSF rhinorrhea.¹⁶⁾

In 1926 Locke⁴⁾ reviewed six reported cases of spontaneous CSF rhinorrhea due to remote tumors and described one of his own. In our review of the literature we found 11 additional case descriptions. All of these cases, as well as our two, are summarized in Table 1. In 13 of these patients, the initial symptoms were signs of tumor; spontaneous CSF rhinorrhea was the presenting symptom in six. Seven patients died of purulent meningitis secondary to CSF rhinorrhea and one succumbed to pulmonary embolism after the second surgery for repair of the CSF leak.⁸⁾ In 12 cases the CSF leaks were successfully repaired: in eight the rhinorrhea stopped after the repair operation; in two it resolved spontaneously

Table 1 Reported cases of spontaneous CSF rhinorrhea as an indirect manifestation of brain tumor

Case No.	Author (Year)	Age	Sex	Type and location of tumor	Site of CSF leak	Initial symptom (duration*)	Outcome (time after last surgery)
1	Nothnagel (1888) ⁷⁾	20	M	tumor of corpora quadrigemina	cribriform plate	tumor signs (4 yrs)	death (< 1 yr)
2	Wollenberg (1899) ⁷⁾	22	M	two occipital lobe tumors	anterior fossa and cribriform plate connected with lateral ventricle	tumor signs (3 yrs)	death (< 1 yr)
3	Meyer (1903) ⁶⁾	20	M	pineal adenoma	funnel-shaped opening from nose to lateral ventricle	tumor signs (5 yrs)	death (< 1 yr)
4	Vigouroux (1908) ¹⁴⁾	28	M	choroid plexus papilloma of fourth ventricle	two funnel-shaped depressions lined with dura and tiny perforations in the frontal base	rhinorrhea	death (some yrs)
5	Souques and Odier (1917) ¹¹⁾	?	?	C-P angle tumor	cribriform plate connected with lateral ventricle	tumor signs (9 yrs)	death (4 days)
6	Cushing (1921) ²⁾	41	M	acoustic neuroma	anterior horn of lateral ventricle communicating directly with nasal cavity	tumor signs (14 yrs)	death (3 yrs)
7	Locke (1926) ⁴⁾	46	F	glioma of C-P angle and frontal lobe	opening through cribriform plate into nose	tumor signs (7 yrs)	death (3 yrs)
8	Love and White (1960) ⁵⁾	22	F	epithelial-lined cyst of fourth ventricle	cribriform plate connected with lateral ventricle	rhinorrhea	no leak after 2nd surgery and drainage (2 yrs)
9	Ommaya <i>et al.</i> (1968) ⁸⁾	?	?	cerebellar glioma	cribriform plate	?	no leak (6 yrs)
10	"	10	M	cerebellar astrocytoma	cribriform plate	tumor signs (11 yrs after tumor resection)	death (3 yrs; pulmonary embolism)

Contd.

following tumor resection; and in two the CSF leak stopped abruptly after shunting procedures.

It is intriguing that CSF leakage can either begin or stop after tumor removal. Our two patients developed CSF rhinorrhea after tumor excision, whereas in the two cases reported by Rovit *et al.*¹⁰⁾ the CSF leaks ceased spontaneously after tumor resection. Our own experiences lead us to speculate that long-standing increased intracranial pressure results in dural defects in the region of the cribriform plate. Subsequently, the fragile cribriform plate may be destroyed, forming fistulas in the process, which are plugged by brain tissue prior to surgery. Removal of the tumor decreases the intracranial pressure, and

the brain tissue then shrinks away from the fistulas, allowing the development of CSF rhinorrhea. In Rovit's two cases spontaneous resolution of the rhinorrhea after tumor removal nullified the necessity for identification of the site(s) of the CSF leak. It may be interpreted that high CSF pressure abruptly dropped after the tumor removal and subsequently the fistula was somehow plugged by surrounding tissue.

The fistula producing the CSF leak was identified at surgery or autopsy in 16 of the 20 cases listed in Table 1. The site most often affected was the cribriform plate (nine cases), followed by the base of frontal fossa (three cases); in two cases both sites

Table 1, page 2

Case No.	Author (Year)	Age	Sex	Type and location of tumor	Site of CSF leak	Initial symptom (duration*)	Outcome (time after last surgery)
11	Ommaya <i>et al.</i> (1968) ⁸⁾	young	F	third ventricle tumor (cranio-pharyngioma?)	unknown (leakage stopped after shunt emplacement)	tumor signs (8 yrs)	no leak (?)
12	Rovit <i>et al.</i> (1969) ¹⁰⁾	41	M	colloid cyst of third ventricle	unknown (leakage stopped after tumor removal)	tumor signs (13 yrs)	no leak (?)
13	"	48	M	choroid plexus papilloma of third ventricle	unknown (leakage stopped after tumor removal)	rhinorrhea	no leak (4 yrs)
14	"	21	M	parieto-occipital meningioma	small protrusion of cortex attached to dural tear in cribriform plate	rhinorrhea	no leak after several episodes of leakage
15	Park <i>et al.</i> (1977) ⁹⁾	32	F	parieto-occipital meningioma	unknown (leakage stopped after shunt emplace-ment)	rhinorrhea	no leak (?)
16	Thinakkal <i>et al.</i> (1980) ¹²⁾	56	M	bilateral C-P angle cholesteatoma	floor of empty sella	rhinorrhea	no leak (?)
17	Vaquero <i>et al.</i> (1981) ¹³⁾	56	F	frontal parasagittal meningioma	meningoencephalocele protruding into sphenoid sinus	tumor signs (1 mo after tumor resection)	no leak (?)
18	von Essen <i>et al.</i> (1981) ¹⁵⁾	46	F	fronto-parietal convexity meningioma	encephalocele protruding into cribriform plate	tumor signs (22 mos after tumor resection)	no leak (?)
19	Present Case 1	49	F	parieto-occipital convexity meningioma	small protrusion of cortex into frontal fossa and cribriform plate	tumor signs (1 mo after tumor resection)	no leak (6 yrs)
20	Present Case 2	37	M	acoustic neurinoma	cribriform plate	tumor signs (4 days after tumor resection)	no leak (6 yrs)

*Interval between appearance of initial tumor sign and first occurrence of rhinorrhea. C-P: cerebellopontine.

were involved. Rare sites of CSF leakage included the floor of an empty sella in the presence of a bilateral cerebellopontine angle cholesteatoma¹²⁾ and the roof of the sphenoid sinus, which contained a meningoencephalocele.¹³⁾ In our two cases, the fistulas were confirmed to be small foramina measuring about 2 mm in diameter. Two were located in the lamina cribrosa and one in the frontal fossa.

A CSF leak that occurs after tumor removal should be repaired as soon as possible because of the unlikelihood of spontaneous resolution and the high potential for the development of purulent meningitis. It should also be noted that CSF rhinorrhea following excision of an acoustic neurinoma often

arises because of the removal of air cells, which is necessary for complete extirpation of the tumor. Therefore, CSF leakage *via* the frontal base should be differentiated from leakage through the eustachian tube.³⁾

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References

- 1) Brisman R, Hughes JEO, Mount LA: Cerebrospinal

- fluid rhinorrhea. *Arch Neurol* 22: 245-252, 1970
- 2) Cushing H: Further concerning the acoustic neuromas. *Laryngoscope* 31: 209-228, 1921
 - 3) Harner SG, Laws ER Jr: Translabyrinthine repair for cerebrospinal fluid otorhinorrhea. *J Neurosurg* 57: 258-261, 1982
 - 4) Locke CE: The spontaneous escape of cerebrospinal fluid through the nose. Its occurrence with brain tumor. *Arch Neurol Psychiat* 15: 307-324, 1926
 - 5) Love JG, White RJ: Cerebrospinal rhinorrhea associated with tumor of the fourth ventricle. Report of a case. *J Neurosurg* 17: 1083-1088, 1960
 - 6) Meyer A: Escape of cerebrospinal fluid through the nose. *J Nerv Ment Dis* 31: 216-217, 1903
 - 7) Nothnagel H: Abfließen von Cerebralflüssigkeit durch die Nase. *Wiener Medizinische Blätter* 11: 161-163, 193-197, 225-228, 1888
 - 8) Ommaya AK, Di Chiro G, Baldwin M, Pennybacker JB: Non-traumatic cerebrospinal fluid rhinorrhoea. *J Neurol Neurosurg Psychiat* 31: 214-225, 1968
 - 9) Park SK, Lee KC, Lee HJ: High pressure cerebrospinal fluid rhinorrhea due to parieto-occipital meningioma. A case report. *Yonsei Med J* 18: 160-165, 1977
 - 10) Rovit RL, Schechter MM, Nelson K: Spontaneous high pressure cerebrospinal rhinorrhea due to lesions obstructing flow of cerebrospinal fluid. *J Neurosurg* 30: 406-412, 1969
 - 11) Souques MM, Odier CH: Ecoulement spontané de liquide céphalorachidien par les fosses nasales, dans un cas de tumeur cérébrale. *Bull Soc Med Hop Paris* 41: 752-761, 1917
 - 12) Thinakkal R, Glista G, Reichman OH, Patel M, Derman G, Borkenhagen R: Rhinorrhea associated with intracranial cholesteatomas and an empty sella. *Laryngoscope* 90: 1679-1685, 1980
 - 13) Vaquero J, Cabezudo JM, Salazar AR, Brasa J: Symptomatic intrasphenoidal meningoencephalocele after removal of a parasagittal meningioma. *Acta Neurochir (Wien)* 57: 61-65, 1981
 - 14) Vigouroux A: Ecoulement de liquide céphalorachidien. Hydrocephalie. Papillome de plexus choroïdes du IV ventricule. *Rev Neurol (Paris)* 14: 281-285, 1908
 - 15) von Essen C, Holmgren E, Nordborg C, Svendsen P: A large convexity meningioma followed by liquorrhea from a nasal encephalocele. A case report. *Acta Neurochir (Wien)* 59: 143-150, 1981
 - 16) Westmore GA, Whittam DE: Cerebrospinal fluid rhinorrhoea and its management. *Br J Surg* 69: 489-492, 1982
 - 17) Wollenberg R: Ein Fall von Hirntumor mit Abfluss von Cerebrospinal Flüssigkeit durch die Nase. *Arch Psychiat Nervenkr* 31: 206-240, 1899

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