Case Report

Untreated Non-Functioning Pituitary Adenoma Causing Spontaneous Cerebrospinal Fluid Rhinorrhea: A Case Report

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Cerebrospinal fluid (CSF) leakage is common with traumatic brain injury or after transsphenoidal surgery (TSS). In contrast, spontaneous rhinorrhea caused by pituitary adenoma (PA) without prior treatment is rather unusual. Moreover, cases of non-functioning PA (NFPA) who seek medical attention without visual or hormonal symptoms but with watery nasal discharge, mistaken for rhinitis, and misled to delayed diagnosis, are extremely rare. The authors presented a case of spontaneous CSF rhinorrhea, confirmed by clinical, laboratory, and radiographic studies, caused by NFPA. Endoscopic TSS came across a typical PA, and subsequently, the sellar defect was successfully repaired. Pathological diagnosis of NFPA was established. The patient had an uncomplicated postoperative course and complete resolution of her presenting symptoms. A short review of prior case reports is also provided.

Keywords: Spontaneous cerebrospinal fluid rhinorrhea, Pituitary adenoma, Non-functioning, Endoscopic transsphenoidal surgery

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Cerebrospinal fluid (CSF) leakage from pituitary adenoma (PA) without an operation is highly unusual. Lam et al reviewed this infrequent condition and found that more than 70%, its most common etiology, is medically treated prolactinoma[1]. However, only seven cases of non-functioning PA (NFPA), without prior treatment, were previously reported for having spontaneous CSF rhinorrhea as their presenting symptom[2-7]. Within the present case information, the authors described a similar patient, the first case in Southeast Asia, who had untreated NFPA with this very rare presentation.

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Case Report

The patient was a 65-year-old woman who had had persistent aqueous rhinorrhea via the left nostril, worse on bending forward, for 14 months. History of head trauma or known neoplastic condition was negative. Diagnosis of rhinitis from other causes was given. Her past visits to various facilities were somewhat inconclusive. As the symptom worsened, she suffered from progressively more debilitating headache associated with intracranial hypotension, for the latest few months. After referral to Ramathibodi Hospital, the rhinorrhea fluid test verified as CSF. Computerized tomography and magnetic resonance imaging scans were obtained (Figure 1, 2). Both modalities demonstrated CSF leakage through a defect at the sellar floor along with a presumed NFPA, based on unremarkable hormonal status. Although the tumor abutted the optic apparatus, her visual acuity was barely affected, measuring at 20/50 on the Snellen chart. However, she was unaware of this acuity change. The visual field, by automated perimetry test, was normal.

The patient underwent an endoscopic trans-

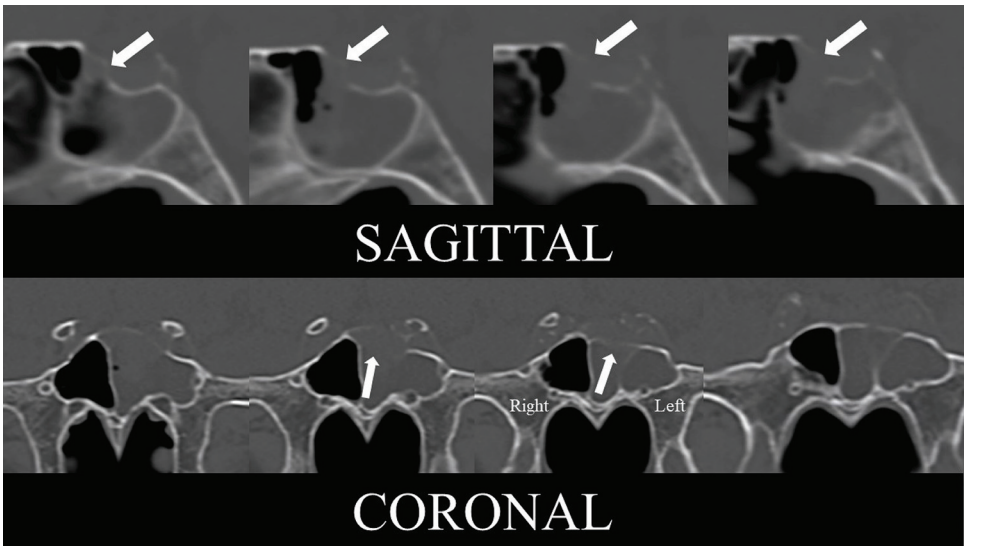

Figure 1. Cranial computerized tomography, sagittal and coronal, scans of the patient illustrating fluid accumulation in the sphenoid sinus without pneumocephalus. White arrows indicating the left-sided sellar floor defect.

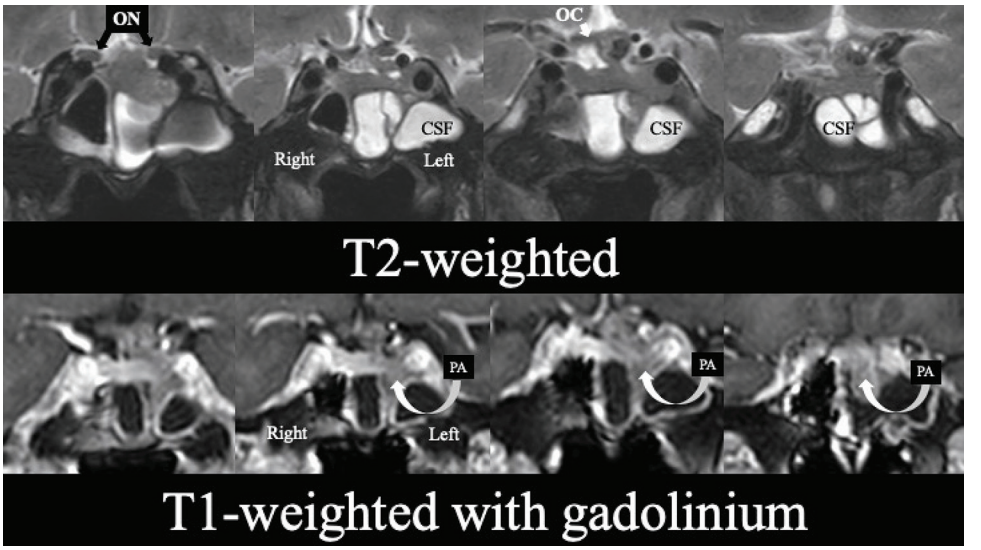

Figure 2. Coronal views of magnetic resonance imaging of the pituitary gland demonstrating hyperintense signal of cerebrospinal fluid (CSF) in T2-weighted images. The curved arrows indicating pituitary adenoma (PA), in gadolinium-enhanced T1-weighted images, at the left side of sella turcica. Note the tumor abutting the optic nerves (ON) and chiasm (OC).

sphenoidal surgery (TSS) that identified the PA, eroding the left-sided sellar floor into the sphenoid sinus. The tumor resection was carried out, followed by multi-layer repair utilizing dura substitute without vascularized nasoseptal flap or lumbar drain. She had an uneventful hospital course, and the final pathology confirmed NFPA. At the last follow-up, the patient had complete resolution of the CSF rhinorrhea and the intracranial hypotension.

Discussion

The most frequent complaints from symptomatic PA are visual and hormonal disturbance. While postoperative CSF leakage is a common complication after TSS for PA, in marked contrast, cases of spontaneous rhinorrhea from PA without prior operation are rare. Within this rare group, most cases were patients who received medication for prolactinomas in which shrinkage of the tumors led

Table 1. Summary of case reports of untreated non-functioning pituitary adenoma with spontaneous CSF rhinorrhea

Author/year	Age (year)/ sex	Duration of rhinorrhea	Meningitis	Pituitary hormone status	Visual acuity/visual field	Surgery	Postoperative CSF rhinorrhea recurrence
Giovanelli, et al.[2]/1967	40/F	6 months	+	Hypo.	Normal/normal	TCS	–
Cole, et al.[3]/1980	28/F	3 months	–	Normal*	Normal/normal	TCS	+
Cole, et al.[3]/1980	38/M	17 months	+	Normal	Normal/normal	TSS	–
Nutkiewicz, et al.[4]/1980	45/F	3 years	–	Partial.	Normal/normal	TCS	–
Rothrock, et al.[5]/1982	24/M	10 years	+	NR	NR	TSS	+
Spaziante, et al.[6]/1991	28/M	9 days	+	Hypo.	Almost blind	TSS	–
Goyal et al.[7]/2012	35/M	6 months	–	Normal	Slightly compromised/normal	TSS	–
Current patient/2020	65/F	14 months	–	Normal	Slightly compromised/normal	TSS	–

F=female; M=male; +=present; –=absent; Hypo.=panhypopituitarism; Partial.=partial hypopituitarism; NR=not reported; TCS=transcranial surgery; TSS=transsphenoidal surgery; CSF=cerebrospinal fluid

* Normal pituitary hormones except secondary amenorrhea from slightly elevated prolactin level

to CSF leakage[1]. The even more rarely encountered entity was the hormone-inactive PA. To date, there were only seven case reports of untreated NFPA with CSF rhinorrhea as the first presentation[2-7].

From the literature search of English language publication for spontaneous CSF rhinorrhea, by excluding case reports without pathological confirmation of chromophobe PA and those with laboratory data indicating hormone-producing PA, such as prolactinoma, Table 1 summarizes the data of the previous seven cases plus the presented patient, who were truly untreated NFPA. In addition, not included in Table 1, were patients who received treatment(s), i.e., medication, surgery, or radiotherapy, before the leakage. With equal gender distribution, most cases were relatively young except the present case. Most had unremarkable visual field with normal, or slightly impaired visual acuity. The only severe visual disturbance at presentation was a 28-year-old man reported by Spaziante et al. Based on their description, he likely suffered from pituitary apoplexy at the first ictus. Nine days later, while improving from initial headache and visual disturbance, the patient developed CSF rhinorrhea and meningitis. Although the duration of his rhinorrhea was not clearly stated, it was assumed to be no longer than nine days[6]. Despite the fact that he did not present with CSF rhinorrhea at the first visit, the authors still included this case into Table 1 for the lack of prior treatment. Except for this mentioned patient, the duration of rhinorrhea was usually long, ranging from 3 months to 10 years[2-5,7]. Because the CSF rhinorrhea was often misdiagnosed as rhinitis due to other etiology, definitive treatment would be delayed. Consequently, four out of eight cases succumbed to meningitis[2,3,5,6]. The present patient was blessed that she did not develop serious infection in spite of protracted CSF leakage. Regarding pituitary function, half of the cases had normal hormones while two patients had panhypopituitarism[2-4,6,7]. Before 1980, surgical repair for the rhinorrhea was often performed by transcranial surgery[2-4]. Recurrent CSF leakage was problematic after surgery in two earlier cases; however, the surgical outcome improved after 1982, as technology evolved, by utilizing TSS[5-7].

In summary, spontaneous CSF rhinorrhea in untreated NFPA is extremely rare. The correct diagnosis mandates complete awareness of this entity. On the contrary, its treatment, once diagnosed, is rather straightforward, by the TSS, as in the present case. Because there was one earlier case report from India[7], the presented patient was, therefore, the second case from Asia. Nevertheless, this is the first case report of such condition in Southeast Asia.

Conclusion

Spontaneous CSF rhinorrhea in an untreated NFPA is extraordinarily rare. Because patients may not have visual or hormonal disturbance, this unusual presentation could be overlooked, and if the correct diagnosis is delayed, meningitis may occur. Nowadays, specific treatment, namely TSS, is highly effective.

What is already known in this topic?

Spontaneous CSF rhinorrhea from PA is uncommon. Most case reports were patients with

medically treated prolactinoma. Untreated NFPA, as the etiology of such condition, is extremely rare.

What this study adds?

Adding to the seven previously described cases worldwide, this is the first report of an untreated NFPA causing spontaneous CSF rhinorrhea from Southeast Asia. Because of delayed diagnosis, lack of definitive treatment would lead to serious complications. Hence, physicians managing rhinorrhea should be aware of this condition.

Conflicts of interest

The authors declare no conflict of interest.

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