Prolactinoma coexisting with cerebrospinal fluid rhinorrhea and cavernous internal carotid aneurysm: Case report and literature review

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ARTICLE INFO

Received: 20 October 2016
Revised: 15 November 2016
Accepted: 10 December 2016

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KEYWORDS

prolactinoma; cerebrospinal fluid rhinorrhea; carotid artery aneurysm; epistaxis

ABSTRACT

Pituitary adenoma coexisting with cerebrospinal fluid (CSF) rhinorrhea and carotid aneurysm is extremely rare. CSF rhinorrhea may cause pneumocephalus and intracranial infection. Rupture of the aneurysm may cause fatal consequence. The authors report such a rare case to draw more attentions. A 55-year-old man presented with sexual dysfunction for 2 years. The serum prolactin was tested as 1,600 ng/ml (normal range, 1.39–24.2). Enhanced cranial MR showed an evident lesion at the sellar area, invading the right cavernous sinus. Prolactinoma was diagnosed. He took bromocriptine for one year and received gamma knife therapy thereafter. Four months after the treatment of gamma knife, he got CSF rhinorrhea and nasal bleeding. The endoscopic transnasal-sphenoidal approach was performed to resect the tumor and repair the dura defect. The CSF rhinorrhea stopped after the surgery, however his nasal bleeding continued. The digital subtraction angiography (DSA) showed an aneurysm at the right cavernous internal carotid. The endovascular coil embolization was performed to treat the aneurysm. The patient recovered well. The coexistence of CSF rhinorrhea and pituitary adenoma is a high risk factor for the rupture of cavernous internal carotid aneurysm. When treating patients with pituitary adenoma and CSF rhinorrhea, doctors should exclude the aneurysm. When nasal bleeding occurs, the hemorrhage of internal carotid should be considered, and appropriate measures should be taken immediately.


1 Introduction

Cerebrospinal fluid (CSF) rhinorrhea is a common complication of patients with pituitary adenoma[1–3]. The invasion of tumor and the operation techniques can destroy the skull base bone and dura. They can cause the communication between the subarachnoid and extracranial space, thus leading to the rhinorrhea[4]. CSF rhinorrhea can cause fatal consequences, such as pneumocephalus and intracranial infection[5].

Cavernous internal carotid aneurysm is a fatal complication of the pituitary adenoma. It may cause serious perioperative or postoperative bleeding[6]. Imamura et al.[7] reported that a patient suffered from pituitary adenoma and cavernous internal carotid aneurysm, he got the aneurysm ruptured and died. Although these cases do not often occur (about 0.2%–1.2%)[8,9], they were described to be associated with significantly high morbidity (24%) and mortality rates (14%)[9]. They should get our attention.

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Supported by the the National Science & Technology Pillar Program during the 12th Five-year Plan Period (No. 2012BAI12B03), Beijing Talents Project (No. 2012D00303400032), National Key Technology Research and Development Program of the Ministry of Science and Technology of China (No. 2013BA09B03), Beijing Institute for Brain Disorders project (No. BIBDPXM2013_014226_07_000084), the National Natural Science Funds for Distinguished 302 Young Scholars (No. 81502390).

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From the year 2008 to 2016, we have treated 36 patients suffering from the coexistence of CSF rhinorrhea and pituitary adenoma. And one of them had got cavernous internal carotid aneurysm. Rupture of the aneurysm can usually cause fatal consequences. Here, we report such a rare case and carefully review the literature.

2 Case presentation

Patient: male, 55 years old, suffered from sexual dysfunction for 2 years. Physical examination showed no positive signs. MRI showed a lesion located at the sellar area, and the tumor invaded the right cavernous sinus (Figure 1a). The serum prolactin was tested as
1,600 ng/ml (normal range, 1.39–24.2 ng/ml). Prolactinoma was diagnosed. And he took bromocriptine for 3 times a day, with a dose of 2.5 mg/time. One year later, cranial MR showed no evident changes in the tumor size, although the level of serum prolactin decreased to 46.3 ng/ml. He then took the gamma knife therapy to inhibit the growth of tumor. Four months after the gamma knife therapy, he got some clear liquid flowing out of the nasal cavity when bowing his head. The liquid was tested, showing Glu: 2.7 mmol/L (range 2.5–4.5 mmol/L). Therefore, the patient was diagnosed with CSF rhinorrhea. After admission, he got the nasal bleeding. The blood amount was about 30 ml, and the bleeding stopped thereafter. At first, the nasal bleeding was ignored. We chose endoscopic transnasal-sphenoidal approach to resect the tumor and repair the dura defect in the diaphragma sellae. The tumor apoplexy was not found during the operation. The dura defect was repaired, and the rhinorrhea stopped after the operation. However, he got another serious epistaxis 3 months after the operation. The bleeding was stopped with stuffing and compression. A suspicious aneurysm of right cavernous internal carotid was found in the MRA (Figure 1b), and the aneurysm was confirmed by the digital subtraction angiography DSA (Figure 1c). The endovascular coil embolization was performed to treat the aneurysm (Figure 1d). After the endovascular treatment, the patient was completely cured and discharged. During the follow-up of next two years, he recovered well and reported no new complaints.

3 Discussion

CSF rhinorrhea and cavernous internal carotid aneurysm can be caused by traumas. And they have also been reported as the complications of pituitary adenoma. There have been many reports of the coexistence of the pituitary adenoma and CSF rhinorrhea[11, 10–17], or pituitary adenoma and aneurysm[18–27]. The incidence of aneurysm in patients with pituitary adenoma has been reported to be 3.7% to 7.4%[28–30]. The mechanism of the coexistence of pituitary adenoma and aneurysm is still unknown. Pia et al.[27] speculated that local circulatory changes of the pituitary adenoma might have induced the aneurysm. Other factors that might cause aneurysms include the changes in collagen metabolism[29], and hormonal influences[29].

Sometimes prolactinomas are invasive. The tumor can destroy the skull base bone and the dura of sellae region, and this can induce CSF leakage. Meanwhile, any significant reduction in the tumor size can also provide a conduit for the escape of CSF, resulting in CSF rhinorrhea[11]. Rapid tumor shrinkage after the dopamine agonist therapy[12], intratumor infarction and/or hemorrhage[15], or increased intracranial pressure can also cause CSF rhinorrhea[31]. When a CSF leak happens, it needs to be repaired.

As the nasal bleeding was not serious before the operation, we didn’t pay enough attention to the symptom. And not until the serious epistaxis happened, no further managements were performed. Once the aneurysm was suspected, we performed angiography examination immediately, and the endovascular treatment was effective. This case taught us a lesson that we should pay attention to every symptom of the patient. Nasal bleeding may be the result of rupture of internal carotid aneurysm in prolactinoma with rhinorrhea. Rupture of aneurysm may be caused by the invasion of the tumor or the operation techniques.

The CSF rhinorrhea in pituitary adenoma patients is mainly caused by the connection between suprasellar cistern and the extracranial space resulting from the invasion of the tumor or the damage of sellar diaphragm dura during the operation. The cavernous segment of internal carotid artery locates near the sellar area, and the average distance of the bilateral internal carotid artery is around 15 mm, with the minimum distance of 9 mm[32]. The cavernous segments of internal carotid artery always locate below the plane of the sellar base, and they often have evident twisting. In the anterior part, the carotid protuberances are located primarily below the sellar base, and are only covered by the dura of the cavernous sinus and the mucosa of the sphenoid sinus in some patients. It means that extensive removal of the lateral sellar floor can be risky[33]. The growths of invasive pituitary adenomas often destroy the bones near the sellar area and the dura of cavernous sinus. Some tumors may grow along the internal carotid artery, and the carotid artery might be damaged by the tumor or the operation techniques. When the aneurysm happens at cavernous segment, it is easily to be injured.

The pituitary adenoma patients who suffer from
preoperative rhinorrhea can be mainly classified into two categories: patients with invasive pituitary adenoma directly destroying skull base structures and patients with macro-prolactinomas after the treatment of bromocriptine. de Lacy et al. reported that pituitary adenoma invading sphenoid sinus often coexisted with CSF rhinorrhea\[34\]. Meanwhile, Raymond et al.\[9\] thought that the susceptible factors causing iatrogenic internal carotid injuries included large invasive pituitary adenomas, trying to remove the cavernous sinus invading tumor through transsphenoidal approach, radiotherapy, and chemotherapy (especially those who take bromocriptine for a long time). The patient in our study had undergone radiotherapy and surgical operation. He had also suffered from the CSF rhinorrhea and epistaxis. We believe that patients with pituitary adenoma and CSF rhinorrhea are more likely to suffer from cavernous internal carotid injury than other pituitary adenoma patients.

MRI has become a routine pre-operative examination for patients with pituitary adenoma. However, MRA and DSA are still not compulsory pre-operative examination for patients with pituitary adenoma. It was reported that patients with pituitary adenoma could suffer from internal carotid aneurysm in cavernous sinus, and the aneurysm could be confirmed by DSA\[35\]. Previously, we didn’t perform angiography examination on patients with pituitary adenoma. But after discovering the present case, we thought that MRA should be performed to know the vascular conditions of the patients with pituitary adenoma and CSF rhinorrhea or patients with internal carotid artery encased by the tumor, especially on the patients with history of nasal bleeding. Even though most patients would get negative results, we thought that this method is meaningful. If positive results are found, it may save the patients’ life.

The rupture of cavernous internal carotid can often cause fatal consequence. Therefore, the possibility of the rupture of aneurysm should be taken into account, before or during the operation, especially in the pituitary adenoma patients with nasal bleeding and CSF rhinorrhea. We should perform emergent vascular examination after controlling the hemorrhage as soon as possible. When dealing with such cases, we used stuffing and compression to stop the hemorrhage. Then the endovascular treatment can be effective.

4 Conclusions

The coexistence of CSF rhinorrhea and pituitary adenoma is a high risk factor for the rupture of cavernous internal carotid aneurysm. When treating patients with pituitary adenoma and CSF rhinorrhea, we should exclude the aneurysm by means of angiography examination. When nasal bleeding occurs, the possibility of hemorrhage of internal carotid should be considered. And appropriate measures should be taken immediately.

References


