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Case Report

Postoperative vasospasm and cerebral infarction in a patient with large pituitary adenoma and cerebral superficial siderosis

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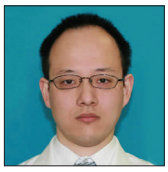

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ABSTRACT

Background: Cerebral vasospasm and infarction are rare complications of transsphenoidal surgery for pituitary adenoma. Cerebral superficial siderosis may result from subarachnoid hemorrhage from a pituitary adenoma. The constellation of cerebral superficial siderosis, cerebral vasospasm, and pituitary adenoma is rare. We describe an extremely rare clinical constellation of immediately postoperative cerebral vasospasm and consequent cerebral infarction in a case with a large pituitary adenoma and cerebral superficial siderosis.

Case Description: A 70-year-old man presented with a pituitary adenoma causing a worsening headache. Preoperative magnetic resonance (MR) images revealed cerebral superficial siderosis, suggesting subarachnoid hemorrhage from pituitary apoplexy. MR angiography (MRA) showed no vasospasm. During the transsphenoidal surgery, an intratumoral hematoma was found. The arachnoid membrane was partially torn and intratumoral hematoma entered the subarachnoid space. Intraoperatively, the intracranial vessels remained intact. The suprasellar tumor was almost entirely resected; however, the patient remained comatose postoperatively. Computed tomography revealed ischemic lesions in the bilateral insular and frontotemporal cortex. MRA revealed cerebral vasospasm in the bilateral middle cerebral arteries. The patient was treated with levetiracetam for nonconvulsive status epilepticus and underwent a lumbar peritoneal shunt surgery for secondary hydrocephalus. However, the patient remained listless.

Conclusion: Postoperative cerebral vasospasm and infarction are severe but rare complications for a pituitary adenoma after transsphenoidal surgery. Preoperative and intraoperative subarachnoid hemorrhage might have been a risk factor in our case. Similar cases should be warranted to analyze whether cerebral superficial siderosis may also indicate the risk of severe postoperative vasospasm immediately after transsphenoidal surgery for pituitary adenoma.

Keywords: Cerebral superficial siderosis, Complication, Ischemic, Pituitary adenoma, Transnasal endoscopic transsphenoidal surgery, Vasospasm

INTRODUCTION

The complication rates related to microscopic and endoscopic transsphenoidal surgery for pituitary adenomas vary from 8.4% to 10.2%. Common postoperative complications include cerebrospinal fluid leakage, cerebrovascular injury, cranial nerve palsy, intracranial hematoma,

meningitis, pituitary insufficiency, and visual impairment.[11] Cerebral vasospasm is a rare complication of transsphenoidal surgery and reports of early symptomatic cerebral vasospasms are scarce.[2,5,11,13] In addition, symptomatic cerebral vasospasm is reported to occur between postoperative days 2 and 16.[2] Several mechanisms possibly causing vasospasm have been proposed such as intraoperative subarachnoid hemorrhage,[1,8] subarachnoid leakage of vasoactive substances from pituitary tumors,[1,8] and postoperative meningitis.[1,8,12,14]

Superficial cerebral siderosis can manifest as hearing impairment, gait ataxia, and cognitive impairment.[6,9,16] Subarachnoid hemorrhage from central nervous system tumors including pituitary adenoma may also result in cerebral superficial siderosis.[9,10,15] However, to the best of our knowledge, concomitant cerebral superficial siderosis, pituitary adenoma, and cerebral vasospasm seem rare. There is no description of postoperative symptomatic cerebral vasospasm in a patient with nonfunctioning pituitary adenoma and cerebral superficial siderosis. Here, we describe a rare case of postoperative symptomatic cerebral vasospasm occurring on the day of the transnasal endoscopic transsphenoidal surgery for a nonfunctioning pituitary adenoma. We also discuss the possible nascent mechanism of this postoperative complication.

CASE PRESENTATION

A 70-year-old man presented with a worsening headache. The patient was suffering from aggravated dementia, diplopia, right dysplasia, and gait disturbance for 6 months. On hospital admission, magnetic resonance imaging (MRI) revealed a tumor in the sella turcica; however, neurological and ophthalmological examinations did not reveal apparent visual loss. The diplopia was confirmed through vertical and horizontal eye movements. The revised Hasegawa dementia scale (HDS-R) and functional independence measure scores were 22 and 109, respectively. Laboratory tests revealed that pituitary hormone was normal. Further MRI revealed homogeneous tumor enhancement, intratumoral hemorrhage, and the tumor extended into the clivus. The cerebrum and cerebellum were atrophic and superficial cerebral siderosis was confirmed in both hemispheres [Figure 1]. Magnetic resonance angiography (MRA) did not reveal cerebral aneurysms, vascular stenoses, or vasospasms and whole-spinal MRI did not reveal any bleeding lesions. In addition, computed tomography (CT) angiography and digital subtraction angiography did not reveal cerebral aneurysms or vascular malformations. Therefore, the preoperative diagnosis was a nonfunctioning pituitary adenoma with apoplexy.

Transnasal endoscopic transsphenoidal surgery

The patient underwent extended transnasal endoscopic transsphenoidal surgery in the supine position under general anesthesia. After preparing a pedicled nasal septal flap and opening the sphenoid sinus, we found a thin wall in the sella turcica and clivus. A dural incision was made and the tumor containing the acute and chronic hematomas was exposed [Figure 2a]. To preserve the pituitary stalk and gland [Figure 2b], the tumor was removed along the margin between the tumor and the arachnoid. During tumor removal, part of the arachnoid was torn due to its adhesion to adjacent nerves and arteries. After nearly total tumor excision, except for the tissue around the nerves or arteries, we confirmed that the arteries were intact. Hemosiderin was observed on the cerebral surface [Figures 2c-e]. The abdominal fat tissue was placed in the removal cavity to prevent cerebrospinal fluid leakage. The sella turcica and clivus dura were closed using the fascia and a nasal septal flap to prevent cerebrospinal fluid leakage.

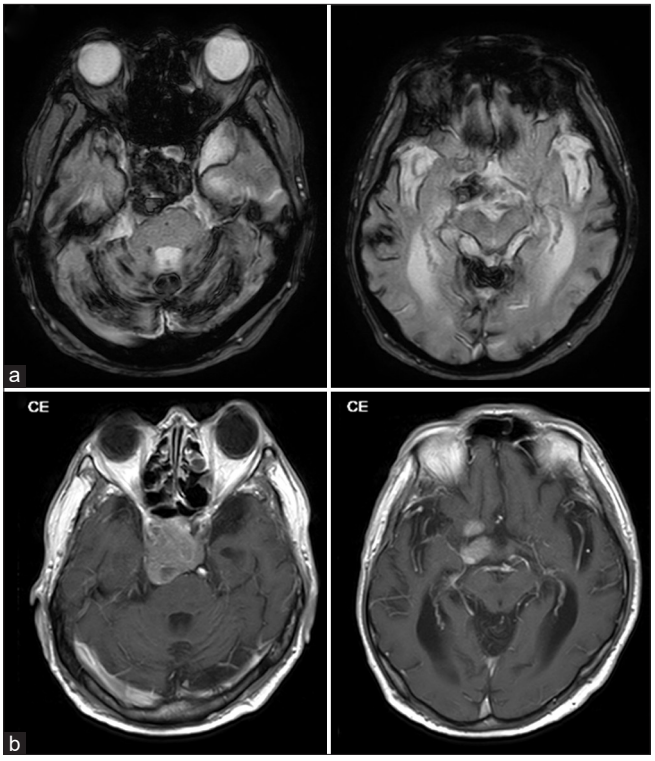

Figure 1: Preoperative magnetic resonance images. (a) Atrophic change observed in the cerebrum and cerebellum. Cerebral superficial siderosis in the bilateral frontal lobes and cerebellar hemispheres, and hemorrhagic change in the tumor were also identified. (b) The tumor in the sella turcica extends close to the brain stem (a: T2 star-weighted images, and b: T1-weighted gadolinium-enhanced images).

Postoperative course

After surgery, the patient remained intubated and unconscious. Postoperative CT imaging on postoperative day 1 showed pneumocephalus and low-density areas in

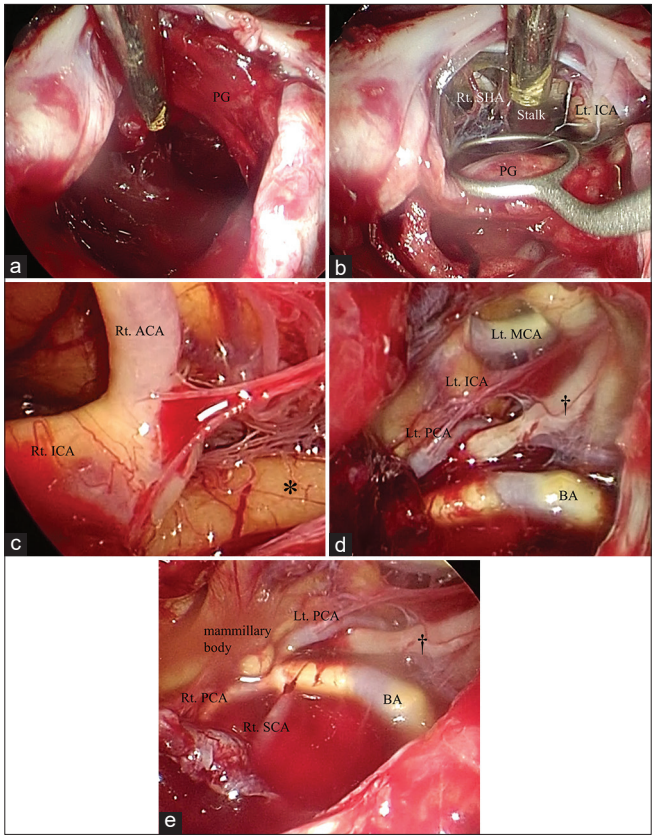

Figure 2: Intraoperative findings under neuroendoscope (a-c, d: under a neuroendoscope angled at 30°, e: under a neuroendoscope angled at 0°). (a) Intratumoral hematoma is observed intraoperatively. The pituitary gland is compressed laterally by the tumor. (b) The pituitary gland and its stalk are preserved intraoperatively. (c-e) After the removal of the tumor in the sella turcica and posterior cranial fossa, the vascular and nerve structures are observed. Intraoperatively, they were preserved. Hemosiderin (asterisk) attaches to the brain surface. The left oculomotor nerve (dagger) is also identified. ACA: Anterior cerebral artery, BA: Basilar artery, ICA: Internal carotid artery, Lt: Left, MCA: Middle cerebral artery, PCA: Posterior cerebral artery, PG: Pituitary gland, Rt: Right, SCA: Superior cerebellar artery, SHA: Superior hypophyseal artery.

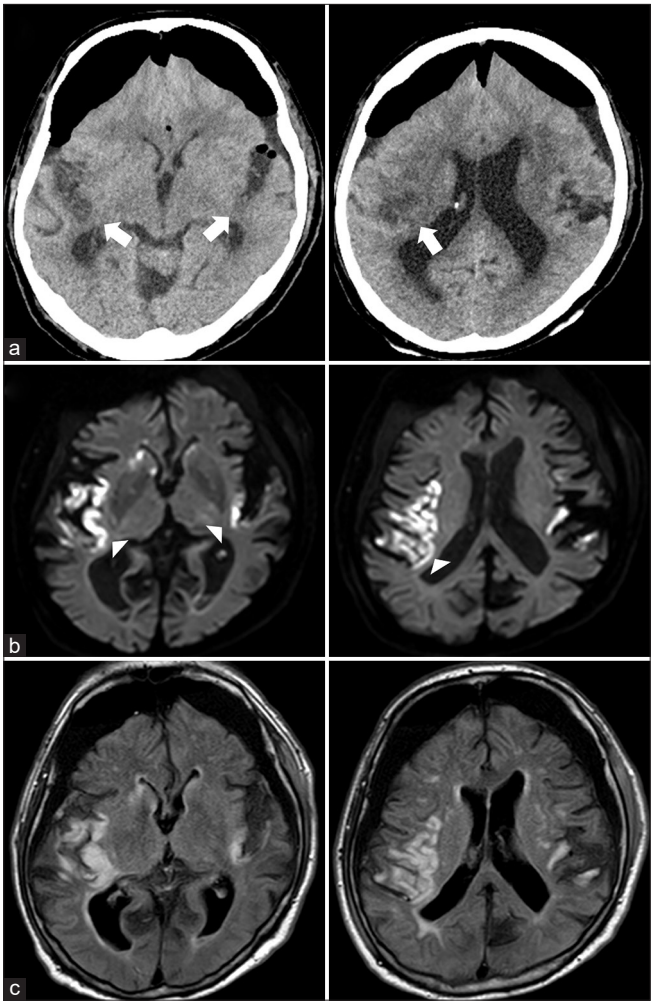

Figure 3: Postoperative radiological images. (a) Computed tomography images on a postoperative day 1 show large pneumocephalus and diffuse ischemic lesions in the bilateral insulae and cerebral cortices (white arrows), (b and c) Acute ischemic lesions in the insula, temporal, and frontal cortices are disclosed bilaterally (white arrow heads). Subarachnoid hemorrhage is not observed. (b) diffusion-weighted images, and (c) fluid-attenuated inversion recovery images.

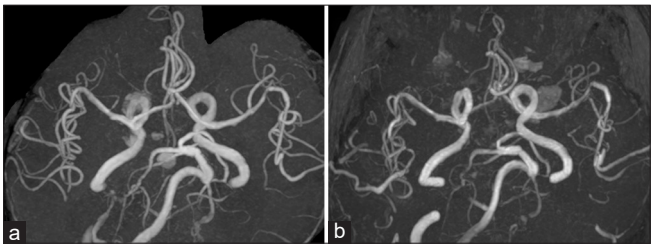

Figure 4: Pre and postoperative magnetic resonance angiography. The intracranial main vessel trunk is clearly visualized on preoperative magnetic resonance angiography (a). However, diffuse spastic changes are observed bilaterally in the middle cerebral artery (b).

the bilateral insular and frontotemporal cortices, with no intracranial hemorrhage [Figure 3a]. The patient underwent the CT imaging on the postoperative day 1, 5, and 7. Postoperative pneumocephalus gradually decreased while the low-density areas were visualized. On postoperative day 5, as the patient's respiration became stable, the patient underwent an MRI examination. An MRA examination was performed to identify the cause of the postoperative low-density areas. MRI and MRA revealed acute ischemic lesions in the left thalamus, bilateral insulae, caudate nuclei, and temporal and frontal cortices [Figures 3b and c], with diffuse vasospasm in the bilateral middle cerebral arteries which was not observed preoperatively. The vasospasm was identified also in the distal middle cerebral artery [Figure 4]. Further, CT angiography, cardiac echography, carotid artery sonography, and lower limb sonography did not reveal any embolic

source. The patient was extubated 7 days after surgery, but his impaired consciousness persisted. Electroencephalography revealed continuous generalized delta and theta activity accompanied by spike waves, suggesting nonconvulsive status epilepticus (NCSE). In addition, a gradual increase in the ventricular size indicated hydrocephalus. The patient was administered levetiracetam (3000 mg daily) and underwent lumbar peritoneal shunt surgery. Although the patient was able to communicate, he was listless with persistent severe cognitive impairment (an HDS-R score of 12). The mean functional independence score was 45. Meningitis did not occur postoperatively. Postoperative hypopituitarism such as adenohypophysis and diabetes insipidus did not occur either. The patient underwent a gastrostomy due to insufficient oral intake on postoperative day 49. Following gastrostomy, the patient was transferred to another hospital to continue rehabilitation therapy at postoperative month 4.

DISCUSSION

Here, we describe a case of postoperative cerebral vasospasm and infarction following transsphenoidal surgery for a nonfunctioning pituitary adenoma, concurrently diagnosed with preoperative cerebral superficial siderosis. Postoperatively, the patient remained unconscious, possibly due to cerebral infarctions in the left thalamus and bilateral cerebral hemispheres, including the insular cortices. In addition, consecutive NCSE appeared to have persisted as a state of impaired consciousness in the patient.

Our case is unique because asymptomatic superficial cerebral siderosis was preoperatively identified. Superficial cerebral siderosis can manifest neurological deficits,[6,9,16] and subarachnoid hemorrhage from central nervous system tumors may be a factor for cerebral superficial siderosis.[9] Mapaga and Martinez reported several causes of cerebral superficial siderosis but did not mention pituitary adenoma.[10] Only two cases of pituitary adenoma (prolactinoma) and cerebral superficial siderosis have been previously described.[10,15] Steinberg *et al*. speculated that cerebral superficial siderosis could have been caused by pituitary apoplexy of the prolactinoma.[15] Although the patient of Steinberg *et al*. had visual disturbances, he refused surgical treatment; therefore, only medication for prolactinoma was prescribed.[15] The pituitary adenomas in those two cases were treated medically with no surgical intervention.[10,15] In our case, pituitary apoplexy was observed on preoperative MRI while the intratumoral hematoma was observed intraoperatively. We hypothesized that the superficial cerebral siderosis may have resulted from minor leakage due to pituitary apoplexy. This hypothesis seems coherent because the patient's history indicated an aggravated headache. Symptomatic cerebral vasospasm usually can occur 2 days after the surgery.[2] Given that the postoperative cerebral spasm occurred on the same day of the surgery, the presence of superficial cerebral siderosis may predispose patients to cerebral vasospasm and infarction during the early postoperative period. Superficial cerebral siderosis can also be observed in probable or possible cerebral amyloid angiopathy.[4] Cerebral amyloid angiopathy is considered a risk for reversible cerebral vasoconstriction.[3] However, it seems controversial whether our case fulfilled the criteria of probable or possible cerebral amyloid angiopathy because of the concurrent pituitary adenoma.[4]

As for the pathophysiology of postoperative cerebral vasospasm in our case, several mechanisms seem involved. Considering the patient's age of 70, surgical manipulation stress, prolonged intraoperative hypotension, and underlying atherosclerosis could have also facilitated the occurrence of postoperative cerebral vasospasm. In our case, the arachnoid membrane was partially torn, when the tumor was surgically detached from the surrounding structures. Although no apparent subarachnoid hemorrhage was observed on postoperative radiological images, minor intraoperative hemorrhagic inflow into the subarachnoid space could have been a cause of the symptomatic cerebral vasospasm in our case.[8] Minor subarachnoid hemorrhage intraoperatively distributed according to the cerebrospinal fluid flow could have led to cerebral vasospasm in the proximal and distal middle cerebral arteries.

NCSE likely results in prolonged unconsciousness. Since cerebral superficial siderosis and pneumocephalus are considered risk factors for NCSE, the concurrent pathology in our case may have resulted in an unfavorable outcome.[7,17] As postoperative pituitary hormone insufficiency did not occur, intraoperative stress of the pituitary stalk seems irrelevant in our case.

To the best of our knowledge, this is the first reported case of cerebral vasospasm and infarction after transsphenoidal surgery for a pituitary tumor presenting concurrently with superficial cerebral siderosis. However, the correlation between these pathologies has not yet been elucidated. Therefore, attention should be paid to the possible risk of postoperative cerebral spastic and ischemic complications after transsphenoidal surgery for pituitary tumors with concurrent cerebral superficial siderosis. Further studies are required to confirm this hypothesis.

CONCLUSION

We describe a rare case of postoperative cerebral vasospasm resulting in ischemic complications after transsphenoidal surgery for a nonfunctioning pituitary adenoma concurrently diagnosed with preoperative cerebral superficial siderosis. Similar reports should be addressed to clarify the perioperative correlation between preoperative superficial

cerebral siderosis and postoperative cerebral vasospasm after transsphenoidal surgery for pituitary adenoma.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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