Simultaneous Occurrence of Subarachnoid Hemorrhage and Epistaxis Due to Ruptured Petrous Internal Carotid Artery Aneurysm: Association With Transsphenoidal Surgery and Radiation Therapy

—Case Report—

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Abstract

A 62-year-old woman presented with simultaneous subarachnoid hemorrhage (SAH) and massive epistaxis. The patient had been treated for pituitary prolactinoma by two transsphenoidal surgeries, gamma knife radiosurgery, and conventional radiation therapy since age 43 years. Cerebral angiography showed left petrous internal carotid artery (ICA) aneurysm with slight stenosis on the adjacent left petrous ICA. She underwent superficial temporal artery-middle cerebral artery (STA-MCA) double anastomosis with endovascular internal trapping without complication the day after onset. Postoperative course was uneventful; the patient did not develop symptomatic vasospasm, recurrent epistaxis, or cerebrospinal fluid rhinorrhea. Postoperative angiography demonstrated complete disappearance of the aneurysm with patent STA-MCA anastomosis. The patient was discharged 2 months after surgery without neurological deficit. The present case is extremely rare with simultaneous onset of SAH and epistaxis caused by ruptured petrous ICA aneurysm. The transsphenoidal surgeries and radiation therapies might have been critical in the formation of the petrous ICA aneurysm.

Key words: cerebral aneurysm, epistaxis, radiation therapy, subarachnoid hemorrhage, transsphenoidal surgery

Introduction

Aneurysms arising at the petrous internal carotid artery (ICA) are rare, and their pathogenesis remains unclear. Petrous ICA aneurysms include pseudo-aneurysms caused by surgical manipulation, radiation therapy, trauma, mycotic, as well as true aneurysms.^{1,2,4,6,10,12)} These aneurysms are usually asymptomatic, but may manifest as hemorrhage or mass effect on the adjacent structures, and can develop variety of clinical signs and symptoms, such as epistaxis, otorrhagia, cranial nerve deficit, vertigo, and dizziness,⁹⁾ whereas association with subarachnoid hemorrhage (SAH) has not been reported. Here, we report an extremely rare case of ruptured petrous ICA aneurysm presenting with simultaneous occurrence of SAH and massive epistaxis.

Case Report

A 62-year-old woman was referred to our institution for the treatment of pituitary prolactinoma in 1990 at the age of 43 years. The tumor was refractory to medical therapy and required surgical management. On initial admission, the huge pituitary tumor was found to extend into sphenoid sinus from the sella turcica with marked erosion of the adjacent bony structure at the skull base. Posterior extension had eroded the dorsum sellae with slight compression of the brain stem, and lateral extension had invaded the left cavernous sinus. The tumor was partially resected via the transsphenoidal approach without removal of the lateral or posterior extensions. The carotid arteries were not injured throughout the surgery. The left intracavernous tumor had gradually grown after initial surgery, which was treated by gamma knife radiosurgery in 1992 (Fig. 1A). The left ICA and surrounding structure received a dose ranging from 25 to 35 Gy (Fig. 1B). However, the

Fig. 1 A: Coronal T₁-weighted magnetic resonance image with gadolinium before gamma knife surgery in 1992 showing the tumor surrounding the left internal carotid artery (ICA). B: Gamma knife planning showing that the left ICA and the surrounding structure received a dose ranged from 25 to 35 Gy. C: Preoperative left internal carotid angiogram in 1996 showing no abnormality.

rest of the tumor outside the radiation field continued to grow. Second transsphenoidal surgery was eventually required to remove the posterior tumor extension which had eroded the clivus with marked compression of the brain stem in 1996. Preoperative angiography revealed no abnormality (Fig. 1C). The carotid arteries were not injured throughout the surgery. She underwent a total of 55.2 Gy of adjuvant radiation following the second surgery with the two opposed portal technique. Magnetic resonance (MR) imaging demonstrated disappearance of the tumor after radiation, and the patient was regularly followed up on an outpatient basis.

The patient was admitted to our hospital with complaints of massive epistaxis in November 2009, 19 years after the initial surgery. She developed severe headache simultaneously with the onset of epistaxis. Hemostasis was achieved by nasal tampon. Computed tomography (CT) revealed SAH with slight hydrocephalus (Fig. 2A). Cerebral angiography showed a left petrous ICA aneurysm with slight stenosis of the adjacent left petrous ICA (Fig. 2B). The upper and lower parts of the aneurysm projected to the cisternal space and the sphenoid sinus, respectively (Fig. 2C). Moreover, coronal CT revealed SAH leaking into the nasal sinus through the skull base defect

Fig. 2 Neuroradiological findings of 2009 hospitalization. A: Initial computed tomography (CT) scans demonstrating subarachnoid hemorrhage (SAH) predominantly located in the basal cistern and empty sella. B: Left internal carotid angiograms, anteroposterior (left) and lateral (right) views, demonstrating the petrous internal carotid artery (ICA) aneurysm and adjacent stenotic segment of the left petrous ICA (arrowheads). C: Coronal T₂-weighted magnetic resonance image showing the aneurysm (arrow) located at the boundary between cisternal space and sphenoid sinus. D: Coronal CT scan showing the SAH leaking into the nasal sinus through the skull base defect (arrowhead).

(Fig. 2D). Balloon test occlusion (BTO) of the left ICA demonstrated the presence of collateral flow via the anterior communicating artery. However, the BTO showed non-synchronous venous filling with reduced regional cerebral oxygen saturation on the occluded side. Therefore, we decided to perform bypass surgery with endovascular internal trapping of the left ICA.

Superficial temporal artery-middle cerebral artery (STA-MCA) double anastomosis was performed via left fronto-temporal craniotomy. The frontal branch and the parietal branch of the STA were anastomosed to the frontal and the temporal cortical arteries (M₄), respectively. Intraoperative angiography confirmed patency of the bypass. Following the bypass surgery, the left ICA including the pseudoaneurysm and the narrowed segment was occluded by

Fig. 3 A: Axial T₂-weighted magnetic resonance images one month after the surgery revealing no ischemic lesions. B: Right internal carotid angiogram, anteroposterior view (left) and left common carotid angiogram, lateral view (right) showing the patent superficial temporal artery-middle cerebral artery double bypass and disappearance of the aneurysm.

endovascular coil embolization in the same operating room. Cerebral angiography after the endovascular treatment showed complete disappearance of the pseudoaneurysm with the patent STA-MCA double bypass.

The postoperative course was uneventful. The patient did not develop symptomatic vasospasm following SAH, recurrent epistaxis, or cerebrospinal fluid rhinorrhea. Postoperative MR imaging and cerebral angiography revealed no ischemic or hemorrhagic complications with the apparently patent STA-MCA bypass (Fig. 3). The patient was discharged 2 months after surgery without neurological deficit.

Discussion

This is a very rare case of petrous ICA aneurysm presenting with simultaneous occurrence of SAH and massive epistaxis. Rupture of petrous ICA aneurysm should not result in SAH because of the extra-arachnoidal localization. In the present case, preoperative coronal MR imaging showed the aneurysm straddled the boundary between the large cisternal space arising in the empty sella and the sphenoid sinus. Furthermore, the skull base bony structure was modified by previous transsphenoidal surgeries, radiation, and tumor invasion. These factors may explain this unusual presentation of simultaneous onset of SAH and epistaxis following aneurysm rupture.

The underlying mechanisms of the aneurysm formation in the present case are unclear. The formation of ICA aneurysms is a rare surgical complication following transsphenoidal surgery.¹¹⁾ Only one case of ICA aneurysm formation was included in a series of 3061

transsphenoidal operations.⁸⁾ ICA aneurysm usually develops in the cavernous portion rather than the petrous portion after transsphenoidal surgery.^{5,7,11)} Rupture of the aneurysm mostly occurs within 2 weeks after surgery, and is rarely delayed.¹¹⁾ These aneurysms are classified as pseudoaneurysm caused by intraoperative direct injury of the arterial wall of the ICA.^{5,7,11)} Another possible pathogenesis of the aneurysm might be radiation therapy. Radiation-induced intracranial aneurysms are rare with only 21 reported cases.^{2,4)} Only 4 cases were petrous ICA aneurysms. Cerebral aneurysms usually take several years to develop after irradiation. Formation of cerebral aneurysm more than 10 years after irradiation has been reported.²⁾ Radiation-induced complications of large cerebral blood vessels are predominantly thrombo-occlusive in nature.³⁾ Aneurysm formation after radiation therapy is less common, and its pathogenesis is still speculative. Accelerated atherosclerosis may be the essential pathogenesis of radiation-induced cerebral aneurysm.³⁾ In the present case, transsphenoidal surgery and radiation therapy might have interacted in the formation of the petrous ICA aneurysm. The petrous ICA may have been manipulated at the second surgery, and was also included within the radiation field. Histological analysis, if possible, might be the key to reveal the pathogenesis of this lesion.

This is an extremely rare case with simultaneous onset of SAH and epistaxis caused by ruptured petrous ICA aneurysm. The previous transsphenoidal surgeries and radiation therapies might have been critical in the formation of the aneurysm and in the rare pattern of onset.

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