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Nontraumatic Internal Carotid Aneurysms in the Paranasal Sinuses Presenting with Epistaxis: A Case Report

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Abstract

Epistaxis due to rupture of a nontraumatic internal carotid artery (ICA) aneurysm in the paranasal sinus has rarely been reported. Here, we report a case of double ICA aneurysms located within both the sphenoid and ethmoid sinuses. A 78-year-old woman presented with recurrent massive epistaxis. Magnetic resonance angiogram (MRA) and cerebral angiogram showed two ICA aneurysms: one protruded into the sphenoid sinus and the other protruded into the ethmoid sinus. Intra-aneurysmal coil embolization was performed for both aneurysms. The patient recovered completely, and a follow-up MRA 3 years later showed no recurrence of the aneurysms. Intra-aneurysmal coil embolization is an option of treatment for an ICA aneurysm filling the paranasal sinus.

Keywords: internal carotid artery aneurysm, sphenoid sinus, ethmoid sinus, epistaxis

Introduction

A total of 16 cases of a nontraumatic internal carotid artery (ICA) aneurysm within the paranasal sinus have been reported to cause epistaxis.[1-16] Among these cases, 15 aneurysms were located in the sphenoid sinus,[1-15] whereas 1 was in the ethmoid sinus.[16] We reported the first case of nontraumatic double ICA aneurysms within the sphenoid and ethmoid sinuses, which were successfully treated with endovascular intra-aneurysmal coil embolization.

Case Report

A 78-year-old woman, who had experienced epistaxis 2 weeks earlier, was taken to an affiliated hospital for a recurrence of severe epistaxis. She had a history of hypertension, chronic kidney disease, and paroxysmal atrial fibrillation treated with apixaban. Moreover, she had a history of repeated severe epistaxis of an unknown cause 20 years ago.

The epistaxis ceased by nasal cavity gauze packing. A few hours later, she had hypotensive shock due to the recurrence of massive epistaxis and was then transferred to our hospital. In the emergency room, transfusion of both fluids and blood and administration of catecholamines improved the shock status; simultaneously, her nasal bleeding was stopped with cramming hemostatic cellulose fiber and gauze into the nasal cavity by an otolaryngologist. She was admitted to the Department of Otolaryngology. Afterward, dialysis was performed due to acute kidney failure caused by the temporary shock. Nasal endoscopy revealed a beating blood clot near the left natural ostium of the sphenoid sinus, which suggested a bleeding point. Blood examination showed anemia and renal failure without coagulation disorders. Head computed tomography (CT) performed 6 days after admission showed opacification of the sphenoid sinus, which suggested the existence of a hematoma, and a partial bone defect in the outer wall of the sphenoid sinus (Fig. 1A). Magnetic resonance angiogram (MRA) performed 7 days after admission showed a small bulge on the cavernous portion of the left ICA into the sphenoid sinus (Fig. 1B), and magnetic resonance imaging (MRI)/T2-weighted volume isotropic turbo spin-echo acquisition demonstrated a low-intensity area that filled the left sphenoid sinus, which indicated a thrombosed aneurysm (Fig. 1C). Moreover, MRA showed another aneurysm with a maximum diameter of 4 mm in the distal cavernous portion of the right ICA. The patient was referred to the Neurosurgical

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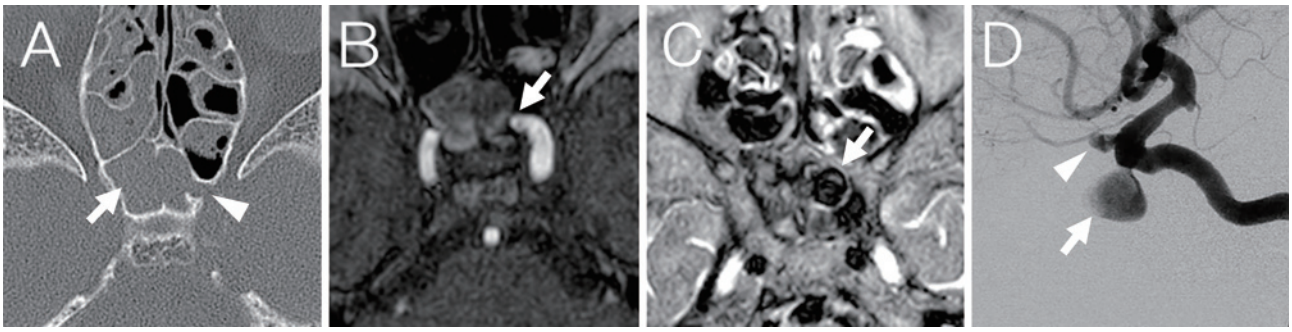

Fig. 1 Preoperative images.
(A) Computed tomogram showing an opacification of the sphenoid sinus (an arrow), which suggests an existence of a hematoma, and a partial bone defect in the left wall of the sphenoid sinus (an arrowhead). (B) Magnetic resonance angiogram showing a small bulge on the cavernous portion of the left internal carotid artery (ICA) (an arrow). (C) A magnetic resonance image/T2-weighted volume isotropic turbo spin-echo acquisition showing a low-intensity area that filled the left sphenoid sinus, which indicated a thrombosed aneurysm (an arrow). (D) Two-dimensional digital subtraction angiogram of the left ICA showing an aneurysm from the cavernous portion of the ICA (an arrow) and another aneurysm on the distal cavernous portion of the ICA (an arrowhead).

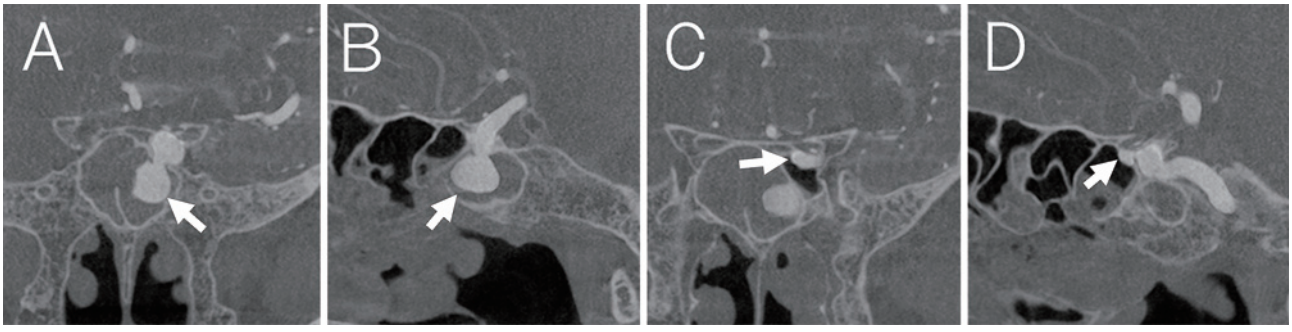

Fig. 2 Fusion images of cone-beam computed tomography and cerebral angiogram.
Coronal (A) and sagittal (B) images showed that the proximal aneurysm was located within the sphenoid sinus and filled the sphenoid sinus with the surrounding thrombosis (an arrow). Coronal (C) and sagittal (D) images showed that the distal aneurysm was located within the ethmoid sinus without surrounding blood clots (an arrow).

Department on the 9th day of hospitalization. Cerebral angiography performed on the same day revealed an aneurysm with a maximum diameter of 11 mm protruding from the cavernous portion of the left ICA into the sphenoid sinus. Another broad-neck ICA aneurysm with a maximum diameter of 7 mm was found on the ICA distal to the first aneurysm (Fig. 1D). As for the crossflow, the right A1 segment of the anterior cerebral artery (ACA) was hypoplastic, and the bilateral ACAs were supplied from the left ICA. A fusion image of a cone-beam CT and cerebral angiogram revealed that the proximal aneurysm was located within the sphenoid sinus and filled the sphenoid sinus with the surrounding thrombosis, which led to the diagnosis of a ruptured aneurysm (Fig. 2A and B). The distal ICA aneurysm protruded into the ethmoid sinus without any surrounding hematomas, which suggested that the distal aneurysm was unruptured (Fig. 2C and D). Since the ruptured aneurysm was surrounded by the bony structure of the sphenoid sinus, using intra-aneurysmal coil embolization, complete occlusion of the aneurysm without recurrence could be expected. To prevent rebleeding, we performed aneurysmal coil embolization the next day under general anesthesia.

A microcatheter was placed in the proximal aneurysm through a catheter in the cervical ICA. Complete occlusion of the ruptured aneurysm was obtained using nine platinum coils with a length of 198.5 mm. Then, the distal ICA aneurysm was occluded by seven platinum coils with a length of 33 mm (Fig. 3A and B).

The patient's postoperative course was uneventful. She was weaned from dialysis due to renal function improvement and was discharged 1 month after admission with modified Rankin Scale score (mRS) of 2 due to her muscle weakness caused by a bedridden state. Three years after discharge, she recovered completely and achieved an mRS score of 0. MRA 3 years after discharge showed complete occlusion of the left ICA aneurysms.

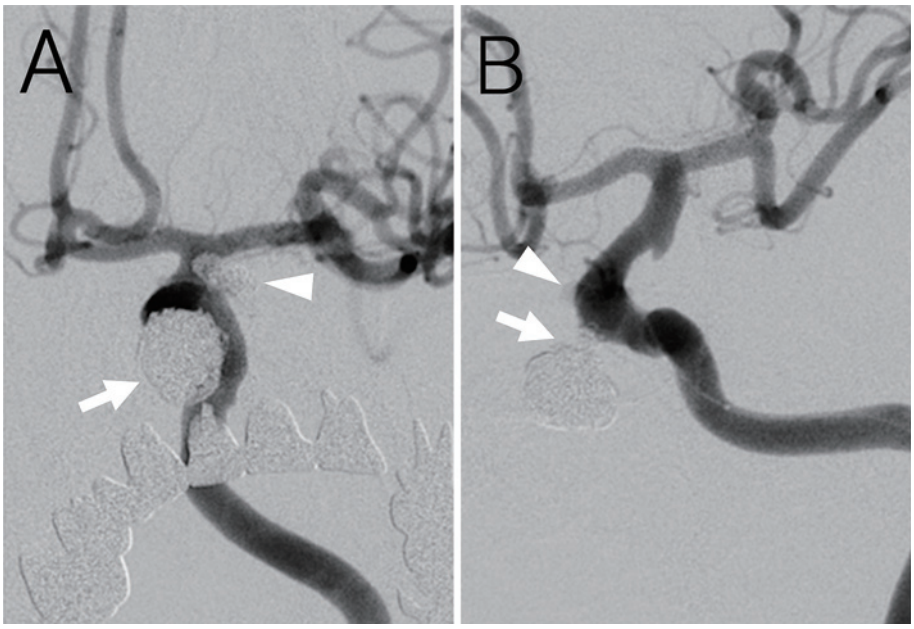

Fig. 3 Postoperative digital subtraction angiogram.
Anteroposterior view (A) and lateral view (B). No blood inflow into the proximal (an arrow) and distal (an arrowhead) aneurysms was shown.

Discussion

We reported a case of epistaxis caused by a ruptured ICA aneurysm in the sphenoid sinus. The most frequent cerebral aneurysm causing epistaxis is a traumatic ICA aneurysm, followed by an infectious aneurysm, and a nontraumatic ICA aneurysm in the cavernous sinus is less frequent.[17,18] A total of 16 cases of ruptured ICA aneurysms in the paranasal sinuses causing epistaxis have been previously reported (Table 1).[1-16] The mean age of the 17 patients, including the patients in this case report (12 women and 5 men), was 62.8 years. The aneurysms were localized in the sphenoid sinus in 16 cases and the ethmoid sinus in 2 cases. To the best of our knowledge, this is the first reported case of double ICA aneurysms, in which one protruded in the sphenoid sinus and the other in the ethmoid sinus.

As for the nature of the aneurysm, a saccular aneurysm was the most probable diagnosis due to the following reasons: the aneurysm showed a narrow neck; parent vessel wall irregularity, which suggested dissecting aneurysm, was not found on a cerebral angiogram; an aneurysm on the contralateral ICA presented; and either an obvious history of paranasal sinusitis or head trauma was not obtained. Therefore, the present aneurysms were possibly neither dissecting aneurysms nor infectious aneurysms.

Anatomically, the ICA prominence exists on the posterolateral wall of the sphenoid sinus. In approximately 2%-23% of the general population, a partial bone defect of the ICA prominence has been reported.[19] All 16 reported aneurysms of the sphenoid sinus extended into the sphenoid sinus through the posterolateral wall bone defect of the sphenoid sinus. A variation of the ethmoid sinus was found in this patient: an ethmoid sinus extending above the sphenoid sinus, which was named Onodi cell,[20] which was observed in approximately 43% of the general population.[21] The previously reported case of an aneurysm in the ethmoid sinus showed the Onodi cell, which was similar to the patient in this case report.[16]

As for the treatment, we chose endovascular coil embolization with preservation of the ICA for the following reasons: the patient's elderly age with renal failure, the aneurysm surrounded by a bony structure of the sphenoid sinus, the narrow aneurysm neck, and a poorly developed crossflow between the bilateral ICAs on a preoperative angiogram. Trapping of the left ICA including the ruptured aneurysm seemed to be invasive, even if the aneurysm ruptured several times. If a sufficient embolization of the aneurysm could not be achieved, additional usage of a stent or ICA trapping with high flow bypass was also planned before treatment.

Until now, seven patients with a paranasal sinus aneurysm, who were treated with intravascular surgery, have been reported.[7-11,13,16] Among them, five were treated using coil embolization alone as the initial treatment[7,10,11,15,16] and two were treated using a combination of coil embolization and stent placement.[8,13] Of the five patients treated with coil embolization alone, only one showed a good outcome,[7] whereas the remaining four showed aneurysm recurrence and subsequently underwent additional treatment, which was a combination of coil embolization and stent placement or flow diverter placement.[10,11,15,16] The pa-

Table 1 Nontraumatic internal carotid aneurysm in the paranasal sinus presenting with epistaxis[1–16)]

Author	Age	Sex	Localization of aneurysm	Treatment	Outcome
Yabe et al., 1987[1)]	70	F	Sphenoid sinus	No treatment	n.a.
Takahashi et al., 1988[2)]	68	F	Sphenoid sinus	ICA ligation + STA-MCA bypass	n.a.
Tamura et al.,1991[3)]	37	F	Sphenoid sinus	ICA ligation after balloon occlusion test	n.a.
Ogawa et al., 1991[4)]	64	F	Sphenoid sinus	ICA ligation + ECA-MCA bypass	Good
Kuba et al., 2000[5)]	36	M	Sphenoid sinus	ICA ligation + STA-MCA bypass	Good
Moro et al., 2003[6)]	64	F	Sphenoid sinus	No treatment	Death
Honeybul et al., 2006[7)]	56	F	Sphenoid sinus	Intra-aneurysmal coil embolization	Good
Lehmann et al., 2009[8)]	41	F	Sphenoid sinus	Intra-aneurysmal coil embolization and stent placement	Good
Nomura et al., 2010[9)]	77	F	Sphenoid sinus	Intra-aneurysmal coil embolization and ICA occlusion after balloon occlusion test	Good
Ronchetti et al., 2013[10)]	81	M	Sphenoid sinus	First, intra-aneurysmal coil embolization; second, intra-aneurysmal coil embolization; third, flow diverter	Good
Akkari et al., 2015[11)]	67	F	Sphenoid sinus	First, intra-aneurysmal coil embolization; second: flow diverter	Good
Garg et al., 2016[12)]	62	M	Sphenoid sinus	ICA ligation + STA-MCA bypass	Good
Sadigh et al., 2016[13)]	47	M	Sphenoid sinus	Intra-aneurysmal coil embolization and stent placement	Death
Kino et al., 2018[16)]	89	F	Ethmoid sinus (Onodi cell)	First, intra-aneurysmal coil embolization; second: intra-aneurysmal coil embolization; third, stent placement	Death
Sriamornrattanakul et al., 2019[14)]	81	M	Sphenoid sinus	ICA ligation + STA-MCA bypass	Good
Grandhi et al., 2019[15)]	50	F	Sphenoid sinus	First, intra-aneurysmal coil embolization; second, flow diverter	Good

ICA, internal carotid artery; ECA, external carotid artery; STA, superficial temporal artery; MCA, middle cerebral artery

tient in this case report is the first reported case of nontraumatic ICA aneurysms in the sphenoid and ethmoid sinuses (Onodi cell), which were treated with intra-aneurysmal coil embolization, showing a good postoperative course.

In conclusion, we reported a case of an ICA aneurysm in the sphenoid sinus presenting with epistaxis, which was successfully treated with intra-aneurysmal coil embolization. Another unruptured aneurysm located in the ethmoid sinus was also embolized subsequently. The patient showed a good outcome without recurrence of the aneurysms.

Conflicts of Interest Disclosure

None.

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