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Technical notes & surgical techniques

Endovascular treatment of a small ruptured non-traumatic internal carotid artery aneurysm presenting with repeated massive epistaxis

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ABSTRACT

Objective: We encountered a rare case of massive epistaxis resulting from a small ruptured non-traumatic cavernous internal carotid artery aneurysm associated a bony defect in the wall of the posterior lateral wall of the sphenoid sinus. We presented successful diagnosis and treatment with a balloon-assisted neck-plasty coil embolization.

Case Presentation: A 74-year-old Japanese man was admitted following recurrent massive epistaxis. There was no history of trauma, surgery, radiotherapy, infection, or tumor. Radiographic imaging demonstrated a bony defect in the wall of the posterior lateral wall of the sphenoid sinus that allowed the aneurysm to ruptured into the nasal cavity rather than intracranially. After rupture, the patient presented with epistaxis that resulted in systemic hypotension. This aneurysm can be managed rapidly using endovascular techniques of a balloon-assisted neck-plasty coil embolization.

Conclusion: The present case report highlights the rarity of a small non-traumatic cavernous ICA aneurysm causing massive epistaxis. Clinicians should be aware of this possibility, as a high index of suspicion is required for proper diagnosis. Internal carotid angiography should be performed, particularly in patients with refractory epistaxis. Aneurysms in this location are usually amenable to endovascular treatment. Coil embolization using a microballoon-assisted neck-plasty technique can result in immediate hemostasis, parent artery preservation, and successful long-term aneurysm occlusion.

1. Introduction

The occurrence of non-traumatic true aneurysms of the cavernous internal carotid artery (ICA) is rare [1], and they represent 3% of all intracranial aneurysms and 11% of all ICA aneurysms [2]. Both endovascular treatment and open surgery play a role in their treatment, but no well-established standard approach exists. Cavernous ICA aneurysms infrequently present with epistaxis; those that do are generally large and associated with radiotherapy, infection, or tumor. Herein, we report the first case of a small (3.5 mm) non-traumatic ICA aneurysm that extended into the sphenoid sinus and ruptured, causing massive epistaxis. The diagnosis, pathophysiology, treatment strategy, and clinical course and outcome are discussed.

2. Case report

2.1. History and examination

A 74-year-old Japanese man with significant epistaxis resulting in hypotension was evaluated by ear, nose, and throat surgeons at another hospital. Surgical nasal cavity exploration and sphenopalatine artery ablation were performed. Although temporary hemostasis was achieved, repeated episodes of epistaxis recurred soon after the procedure. The patient then underwent a nasal mucosa biopsy that revealed no malignancy. Further imaging studies showed a saccular aneurysm of the cavernous segment of the left ICA. At this point, the patient was urgently transferred to our department for definitive treatment.

The patient had a history of diabetes mellitus and asymptomatic hepatitis C virus infection. He denied the presence of hypertension, trauma, antiplatelet or anti-coagulation medication use, and easy bruising or bleeding. He was neurologically intact without the signs of meningismus. Non-contrast computed tomography (CT) demonstrated a bony defect in the left posterior lateral sphenoid sinus wall (Fig. 1A) and blood in the sphenoid sinus without intracranial hemorrhage. A CT angiogram (CTA) demonstrated a 3.5-mm left cavernous ICA aneurysm that protruded into the sphenoid sinus though the bony defect (Fig. 1B). Magnetic resonance imaging showed no evidence of neoplasia around

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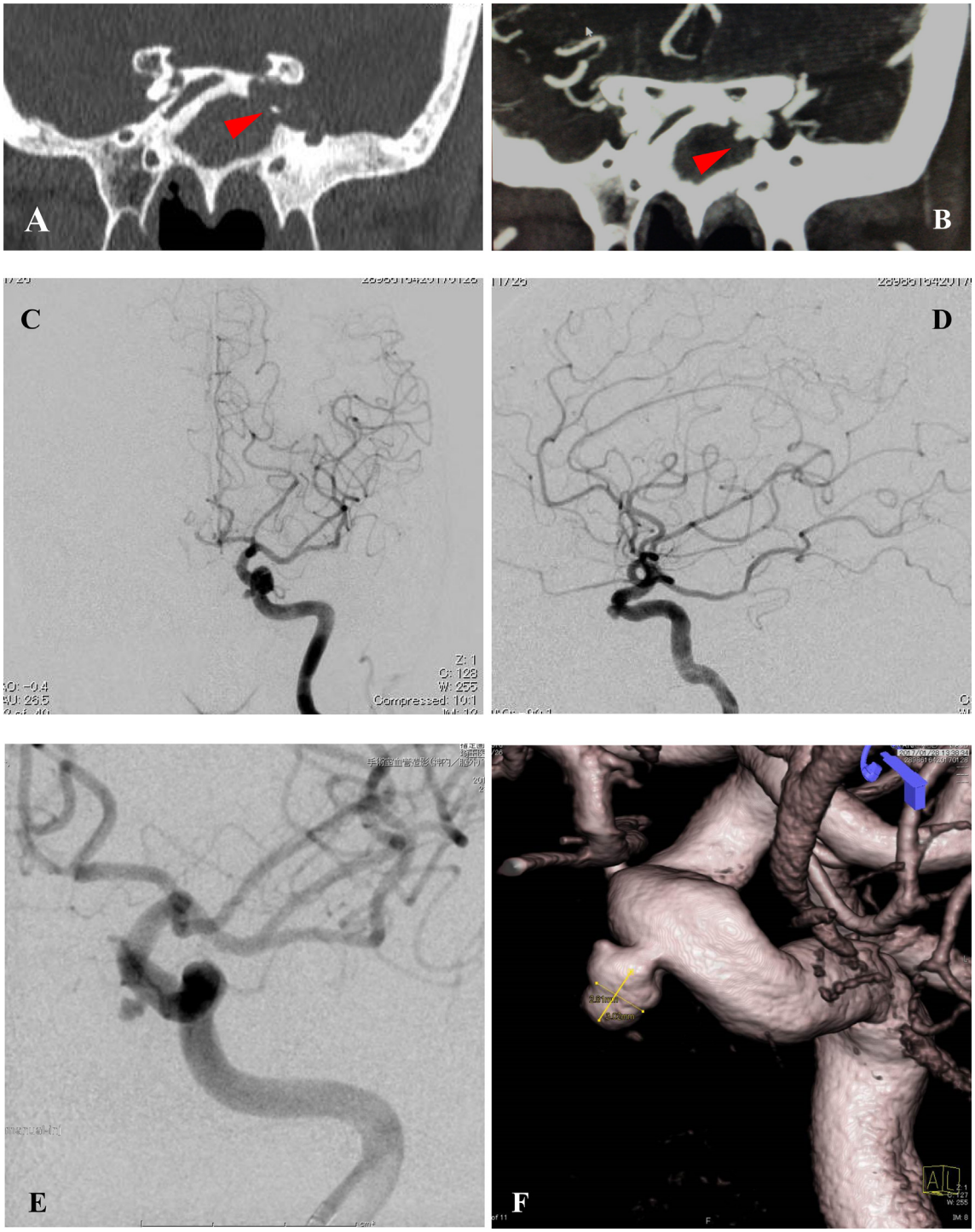

Fig. 1. (A) Computed tomography (CT) scans showing a bone defect in the lateral wall of the left sphenoid sinus (arrowhead). (B) Coronal enhanced CT demonstrating the aneurysm protruding into the sphenoid sinus (arrowhead). (C and D) Anteroposterior and lateral digital subtraction angiography after left internal carotid injection revealing a true aneurysm. (E and F) Initial selective angiography of the left internal carotid artery.

the carotid artery. Emergent digital subtraction angiography was performed and confirmed the irregularly shaped left cavernous ICA aneurysm (Fig. 1C and D).

2.2. Therapeutic course

We considered this as a non-traumatic true saccular aneurysm and decided that endovascular coil embolization was the most feasible treatment for preventing rerupture and preserving blood flow through the parent artery.

After obtaining informed consent, the patient underwent coil embolization of the aneurysm under general anesthesia. At the beginning of the procedure, 2000 IU of heparin was intravenously (IV) administered. Next, a 100-cm 6-French Guiding catheter (Fubuki AN 110 cm, ASAHI INTECC CO., LTD., Aichi, Japan) assisted by a 6-French guiding sheath (AxcelGuide 80 cm, MEDIKIT CO., LTD., Tokyo, Japan) was placed into the left ICA via the right femoral artery. A microcatheter (Neurodeo 10, Medico's Hirata Inc., Tokyo, Japan) was manipulated

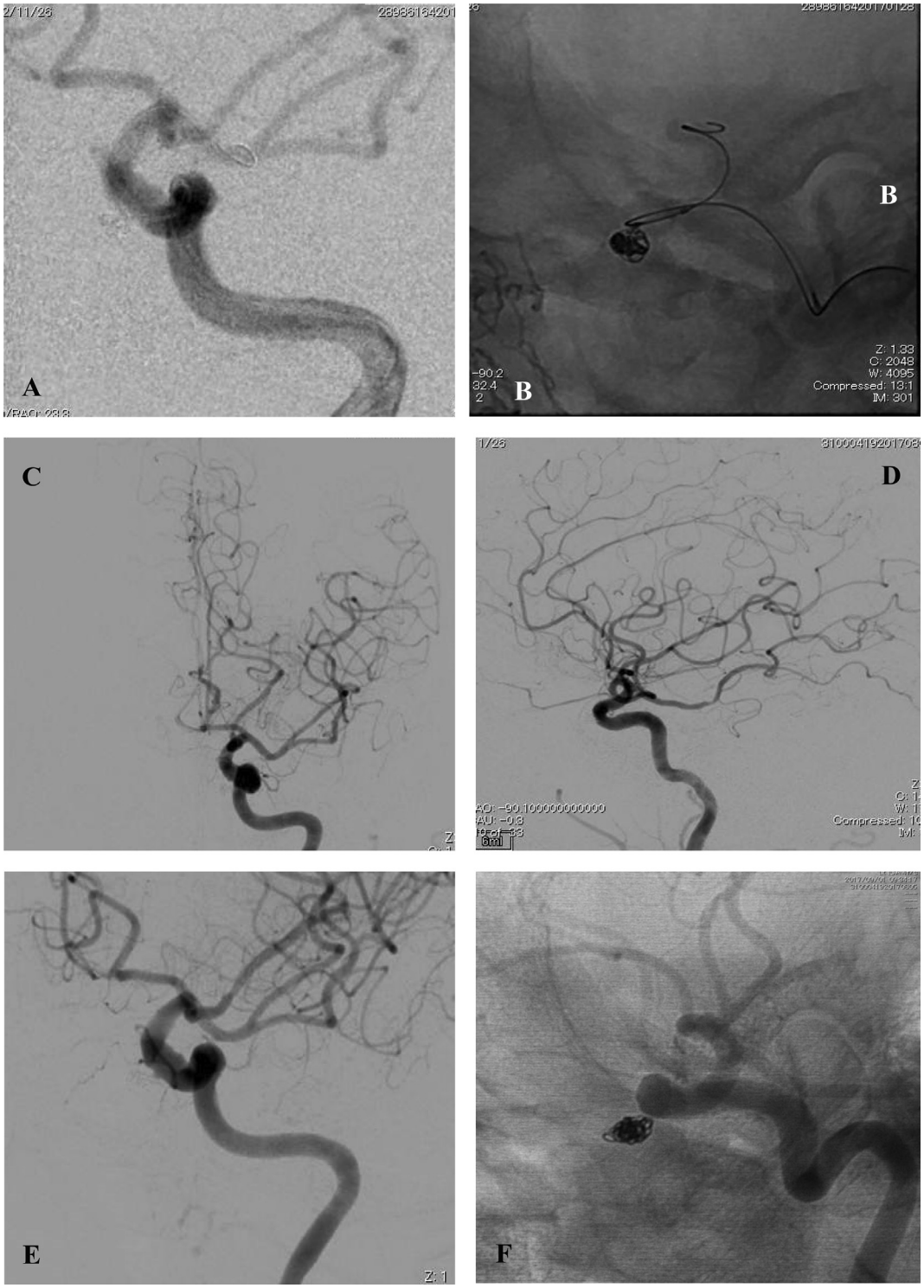

Fig. 2. Selective angiography of the left internal carotid artery following treatment. (A) Subtracted imaging shows complete occlusion of the carotid aneurysm. (B) Unsubtracted imaging. (C and D) Satisfactory packing of the coils and covering of the aneurysm neck at 8-month follow-up. (E) Subtracted imaging shows no revascularization of the aneurysm. (F) Unsubtracted imaging shows no coil migration of material. Preoperative conventional cerebral angiography shows Acom unruptured aneurysm.

into the aneurysm over a microguidewire (Neuroute 14, Medico's Hirata Inc., Tokyo, Japan). The aneurysm was completely occluded with seven coils (one Target 360 Nano 2.5 mm × 4.0 cm, two Target 360 Nano 1.5 mm × 3.0 cm, and four Target 360 Nano 1 mm × 2.0 cm; Stryker, Michigan, U.S.A.) under neck-plasty technique with a micro-balloon (Hyperform 4 mm × 7 mm, eV Neuro-vascular, Irvine, CA, U.S.A.).

Following the procedure, the patient was neurologically intact; nasal packings were removed the following day without any further bleeding. An angiogram 8 months later showed complete obliteration of the aneurysm (Fig. 2A–F). Follow-up magnetic resonance cerebral angiography two years after embolization also showed a stable result. The patient has remained neurologically intact without epistaxis following the treatment.

3. Discussion

3.1. Non-traumatic small internal carotid aneurysm

To the best of our knowledge, this is the first report of a small non-traumatic cerebral aneurysm causing massive epistaxis [3]. Symptoms associated with mass effect usually constitute the primary presentation of non-traumatic cavernous ICA aneurysms (79% of cases) rather than epistaxis (3%) [2]. To date, all reported epistaxis cases have been associated with large ICA aneurysms.

The exact mechanism through which a sterile soft tissue mass erodes through bone remains unknown, and there is a paucity of literature on the topic. However, the osteolytic process associated with bony compression by a large aneurysm has been partially explained by complex molecular interactions that ultimately lead to activation of metalloproteinases and osteoclastogenesis. We hypothesized that the mechanical force from chronic pulsations of the aneurysm against bone leads to bony erosion. Although in our case, the aneurysm was too small to erode the bone.

Fujii et al. conducted an anatomical microsurgical study and found an absence of bone between the carotid artery and sphenoid sinus in 8% of subjects [4]. This absence of bone can allow aneurysm growth into the sinus; subsequent rupture would result in epiastxis. In the present case, this may have played a role, provided the CT finding of a bony defect in the posterior lateral sphenoid sinus wall (Fig. 1). Another possibility is that an unidentified previous sentinel rupture may have occurred and caused secondary local inflammation and bone destruction. Although the exact biological pathway remains unclear, these two mechanisms may have incited molecular cascades that would ultimately result in osteoclastogenesis and bone erosion.

3.2. Aneurysm treatment

Endovascular embolization is an alternative to open surgical treatment for aneurysms of the cavernous, clinoidal, and ophthalmic ICA segments. Its advantages are immediate aneurysm occlusion and hemostasis without effect on the parent vessel. On the basis of our experience with the case presented here, it is possible to perform coil embolization of a small aneurysm with a clear neck using a microballoon-assisted neck-plasty technique without a stent. This stent-free technique reduces the risk of bleeding in the future: there is no need to administer antiplatelet drugs to prevent stent-related thromboembolic complications. However, it is important to carefully assess the shape of the aneurysm to determine if balloon-assisted coil embolization without stent placement is feasible. In the present case, the post-embolization angiogram showed the occluded aneurysm sac and preservation of normal ICA flow.

A recent study conducted on patients who underwent endovascular treatment of ICA aneurysms showed a very low recurrence rate with minimal complications [5]. In carefully selected patients, this approach can successfully treat intracranial aneurysms and negate the need for invasive surgery [5]. Rapid post-procedure recovery and the avoidance of parent artery occlusion and the complications of open surgery are potential advantages of endovascular treatment.

4. Conclusions

The present case report highlights the rarity of a small non-traumatic cavernous ICA aneurysm causing massive epistaxis. Clinicians should be aware of this possibility, as a high index of suspicion is required for proper diagnosis. Internal carotid angiography should be performed, particularly in patients with refractory epistaxis. Aneurysms in this location are usually amenable to endovascular treatment. Coil embolization using a microballoon-assisted neck-plasty technique can result in immediate hemostasis, parent artery preservation, and successful long-term aneurysm occlusion.

5. Sources of funding

None.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.inat.2020.100732.

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