

CASE REPORT

Rupture of Internal carotid artery pseudoaneurysm in the sphenoid sinus as a complication of deep neck space infection

Yoshiyuki Ozono¹, Suetaka Nishiike¹, Masahiro Ishihara², and Toshiyuki Fujinaka³

¹Department of Otorhinolaryngology - Head and Neck Surgery, Osaka Rosai Hospital, Sakai, Japan, ²Department of Neurosurgery, Osaka Rosai Hospital, Sakai, Japan, ³Department of Neurosurgery, Osaka University Graduate School of Medicine, Suita, Japan

Abstract : Background : Pseudoaneurysm of the internal carotid artery (ICA) is a very rare but potentially fatal complication of deep neck space infection. **Methods :** This paper describes a very rare case of an ICA pseudoaneurysm rupture in the sphenoid sinus caused by a deep neck abscess. **Results :** A 62-year-old male with a deep neck space infection underwent surgical drainage. On the postoperative 21st day, however, he suddenly had massive epistaxis. A transnasal endoscopic examination found massive bleeding out of the sphenoid sinus. Immediate intra-arterial angiography revealed two pseudoaneurysms of the left ICA at the cavernous segment (C4) and the clinoid segment (C5), which were embolized with coils. The patient made an uneventful recovery after the embolization. **Conclusion :** We found no reports in the literature that pseudoaneurysms associated with a deep neck infection rupture in the sphenoid sinus. Prompt treatment along with accurate diagnosis is essential for successful management of such cases. *J. Med. Invest.* 66 : 188-189, February, 2019

Keywords : Aneurysm, endovascular procedures, epistaxis, internal carotid artery

INTRODUCTION

Pseudoaneurysm of the internal carotid artery (ICA) is a rare complication of a deep neck infection(1). The rupture of an ICA pseudoaneurysm is life-threatening, and difficult to manage. Here we describe a rare case of a deep neck abscess presenting with massive epistaxis from ICA pseudoaneurysms, which were successfully treated with endovascular embolization. This is the first case report in the world literature of an ICA pseudoaneurysm rupture in the sphenoid sinus caused by a deep neck abscess(1-3).

CASE REPORT

A 62-year-old man who complained of fever, fatigue and left neck pain was referred to our emergency department. His medical history revealed no obvious abnormalities. His left neck was swelling. A CT revealed a deep neck abscess (Fig 1A). It extended to the skull base, but there was no aneurysm at that time (Fig 1B). He was admitted to the Otorhinolaryngology-Head and Neck Department, underwent external drainage, and was given intravenous antibiotics. Intraoperative cultures grew *Streptococcus constellatus*.

On the postoperative day 21, he had a sudden massive epistaxis, and his hemoglobin level decreased to 8.5 g/dl. We performed an endoscopic examination under general anesthesia, and found coagulum in the natural ostium of the sphenoid sinus. While removing it, massive epistaxis recurred from the sinus. We performed posterior nasal packing and consulted the Department of

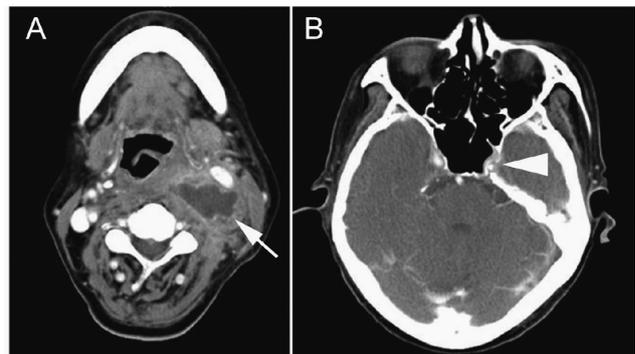


Fig. 1 Contrast-enhanced CT image of the first examination. A : low-density, ring-enhancing mass (arrow) spread to the left carotid sheath. B : No aneurysm was seen in the sphenoid sinus (arrow head).

Neurosurgery.

An intra-arterial angiogram revealed two large saccular aneurysms at the cavernous segment (C4) and the clinoid segment (C5) of the ICA(4) (Fig 2A). They were treated with coil embolization. Post-embolization angiogram showed complete occlusion of the left ICA (Fig 2B). The patient made an uneventful recovery, and at 1-year follow-up, no neurological sequelae were evident.

DISCUSSION

Epistaxis from the rupture of an ICA pseudoaneurysm is extremely rare, and recent reports have been only children cases(1-3). In the past cases, all the pseudoaneurysms were identified in the parapharyngeal space, but, in the present case, we found ICA pseudoaneurysms at the clinoid segment (C5) and the cavernous

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Address correspondence and reprint requests to Suetaka Nishiike, Department of Otorhinolaryngology- Head & Neck Surgery, Osaka Rosai Hospital, 1179-3 Nagasonecho, Kita-ku, Sakai, Osaka 591-8025, Japan and Fax : +81-72-255-3349.

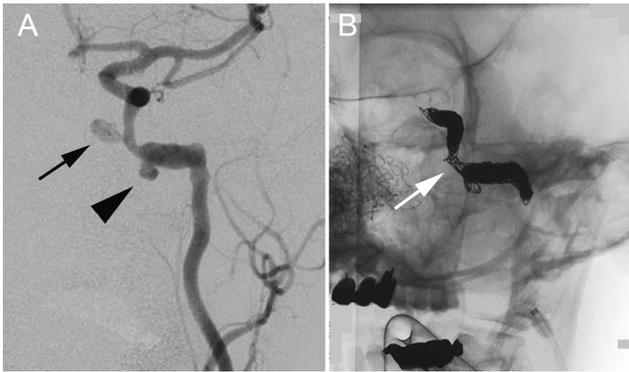


Fig. 2 Angiogram. A : Two large saccular aneurysms with an irregular outline arise from the supposed cavernous segment (C4) (arrow head) and clinoid segment (C5) (arrow) of the left ICA. B : The left ICA was completely occluded with coil embolization (arrow).

segment (C4) (4). We found no reports in the literature that pseudoaneurysms at the clinoid segment associated with a deep neck infection rupture in the sphenoid sinus.

In this case, from the abscess of the patient, intraoperative cultures grew *Streptococcus constellatus*, which is referred to as the *Streptococcus milleri* group (SMG). The SMG is common in the mouth and can be an aggressive pathogen and cause abscess formation(5). SMG infection produces a tissue-destroying enzyme and immunosuppressive substances. And as a result, the infection spreads rapidly and leads to complications at a high rate(6, 7). This may be why this abscess extended to the skull base and created pseudoaneurysms in this case.

Deep space infections most commonly result from otolaryngological or upper respiratory tract infections in children and odontogenic infections in adults(8). The patient did not have such a medical history. As noted above, however, the culture result suggests that this patient's abscess originates from odontogenic infections.

This patient had an interval of 22 days between the surgical drainage for deep neck infection and the diagnosis of pseudoaneurysm of the ICA. The interval between the initial infection and the diagnosis of pseudoaneurysm is as long as 2 months in cases of deep neck infections(9). Previous reports also had a long delay until the diagnosis because of no signs of pseudoaneurysm before epistaxis(1-3).

Physicians who meet such a case would face a dilemma to select a choice between unfavorable treatment options. An endoscopic suture of the ICA rupture has been reported previously(10), but the procedure while confronting of massive bleeding may be severely difficult. An ICA sacrifice such as a ligation of the ICA at the cervical segment or selective arterial embolization of the ICA would be adequately conceivable. The latter option, which can be performed continuously after diagnostic intra-arterial angiography, would be the most realistic countermeasure against the rupture of the ICA pseudoaneurysm in the sphenoid sinus.

In our case, the patient was treated with coiling of ICA. Two of the similar past three cases(1, 2) were also treated with coiling, and the other was with stent(3). It has been reported that ICA permanent emboli are a useful and sometimes necessary procedure for

the treatment of large aneurysms(11). But there is a big risk of stroke with abrupt ICA sacrifice. Fortunately, as a sequel to the emergent ICA embolization, the patient developed neither major nor minor complications.

CONCLUSION

This case suggests that when a patient complains of unknown epistaxis after deep neck infection, physicians should rule out the existence of a pseudoaneurysm complicating deep neck infection, and examine the nose and throat intensively. Prompt treatment along with accurate diagnosis is essential for successful management of such cases.

CONFLICT OF INTEREST

The authors declare that they have no conflicts of interest.

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