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Case Report

Ruptured infectious ICA pseudoaneurysm into the sphenoid sinus after maxillofacial infection, successfully treated by selective embolization

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ABSTRACT

Background: Intracranial infectious aneurysms are cerebral aneurysms caused by pathogen-induced inflammation undermining the arterial wall. We present a rare case of inflammatory pseudoaneurysm of cavernous internal carotid artery (ICA).

Case Description: A 51-year-old female with a recent diagnosis of acute lymphoblastic leukemia developed maxillofacial infection with *Pseudomonas* and *Acinetobacter* after chemotherapy onset. Initial plain computed tomography (CT) revealed bony dehiscence of the left ICA canal, as well as bilateral protrusion of the vessel within the sphenoid sinus. Following infection spread into the left sphenoid sinus, she presented with episodes of intermittent epistaxis, without any profound vascular abnormalities on postcontrast CT. CT angiography that was performed 15 days later, due to refractory epistaxis, illustrated a large narrow necked irregular shape pseudoaneurysm of the left paraophthalmic ICA, extending into the ipsilateral sphenoid sinus. The aneurysm was completely occluded by selective embolization without parent or adjacent vessel sacrifice, documented on both intraoperative and follow-up angiogram, with no recurrence of epistaxis.

Conclusion: Conclusively, ruptured internal carotid infectious aneurysms are rare but potentially fatal causes of epistaxis when extended into the sphenoid sinus. Selective coiling is feasible and can provide definitive treatment of these lesions.

Keywords: Coiling, Epistaxis, Infectious pseudoaneurysm, Maxillofacial infection, Sphenoid sinus

INTRODUCTION

Intracranial infectious aneurysms (traditionally referred to as mycotic aneurysms) are cerebral aneurysms caused by pathogen-induced inflammation undermining the arterial wall. They are relatively rare, accounting for 2.6–6% of all cerebral aneurysms, and are associated with high risk of rupture and mortality.[2] They most commonly arise by hematogenous seeding of systemically circulating microorganisms (often on an already formed atherosclerotic vessel) but may also arise by invasion from adjacent infected tissue. Infectious aneurysms of the internal

carotid artery (ICA) as a result of spreading of paranasal sinus infections are rarely described in the literature, usually on immunosuppressed patients and almost exclusively as a result of fungal infections.[3] Treatment options include a combination of an extensive antimicrobial regiment, endovascular coiling – often with parent vessel sacrifice – and/or stenting, or bypass surgery.[3]

We present a rare case of a left paraophthalmic ICA infectious pseudoaneurysm invading the sphenoid sinus which presented with refractory epistaxis, resulting from a multidrug-resistant *Pseudomonas* and *Acinetobacter* infection and treated successfully by selective coiling with parent vessel preservation.

CASE REPORT

A 51-year-old female with a recent diagnosis of acute lymphocytic leukemia was admitted to our hospital due to persistent fever and left-sided perioral dysesthesia following initiation of chemotherapy. Clinical examination revealed ulcerous lesions in the patient's palate. Further laboratory tests were notable for pancytopenia and elevated CRP and ESR. Cultures of tissue samples taken from the oral lesions revealed multidrug-resistant *Pseudomonas aeruginosa* and *Pseudomonas fluorescens* and intermediate antibiotic sensitivity *Acinetobacter baumannii*. Chemotherapy was discontinued and the already instituted empiric antibiotic coverage was adjusted according to antibiograms. Head and facial computed tomography (CT) revealed extensive bone erosion of the maxilla around the dental alveoli without signs of inflammation of the paranasal sinuses. Anatomic variation was the bony dehiscence of the superolateral wall of the left sphenoid sinus [Figure 1a], with concomitant carotid artery protrusion into the sinus and separated by the sinus cavity by only dura and mucosa.[1,4] One and a half month later, there was sudden-onset epistaxis that was managed successfully by tamponade by ENT surgeons. Postcontrast head and facial CT imaging revealed inflammation of the left nasal cavity and paranasal sinuses –especially the left sphenoid sinus – without any gross vascular abnormalities. Further episodes of intermittent epistaxis were noted although clinical and laboratory signs of infection remitted during the following fortnight. A repeat head-facial contrast CT scan was performed at that time. It showed containment of the infection but revealed a large enhancing lesion protruding into the left sphenoid sinus, which on further CT angiography (CTA) and digital subtraction angiography (DSA) were determined to be a 0.9 × 2 cm infectious irregular multilobulated pseudoaneurysm of the cavernous ICA [Figure 1b, c and 2a]. The patient underwent emergent complete selective endovascular occlusion of the pseudoaneurysm with coils. Stenting of the parent ICA was not necessary and contraindicated in this acute phase, due to the need for concomitant dual antiplatelet therapy. Stenting would have raised concerns for recurrent hemorrhage, compounded by the fact that the patient was possibly due to become thrombocytopenic due to planned continuation of anti-leukemic chemotherapy. Postembolization angiogram revealed complete exclusion of the pseudoaneurysm with preservation of normal flow in the distal carotid and ophthalmic artery [Figure 2b]. There were no postoperative complications, complete remission of epistaxis was documented, and the patient was fully mobilized on the 1st postoperative day. Follow-up CTA a week after embolization confirmed complete occlusion of the aneurysm with no extension of the aneurysm neck or residual flow [Figure 1d].

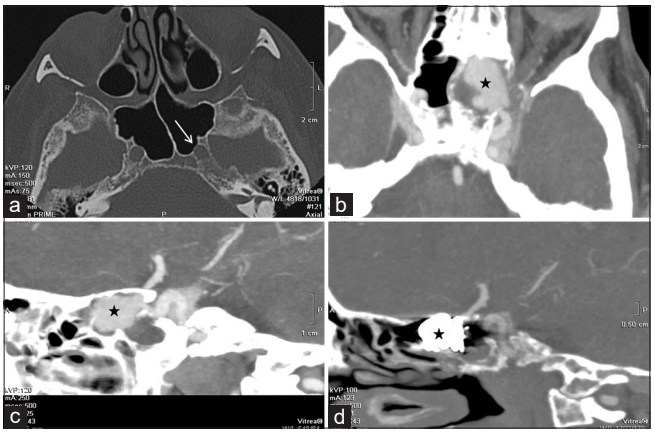

Figure 1: (a) Bone window computed tomography (CT) demonstrating bony dehiscence of the superolateral wall of the left sphenoid sinus (white arrow). (b and c) Preoperative axial (b) and sagittal reconstruction (c) CT angiography (CTA) showing protrusion of the internal carotid artery (ICA) infectious aneurysm into the left sphenoid sinus (asterisk). (d) Postoperative sagittal CTA demonstrating complete occlusion of the aneurysm (asterisk) and good patency of the supraclinoid ICA.

DISCUSSION

Whereas usually benign, sphenoid sinus infection has the potential to cause life-threatening complications in the form of ruptured infectious intracranial aneurysms of the ICA, especially in immunocompromised individuals.[2] A predisposing factor seems to be an anatomic variation in the form of bony dehiscence of the superolateral wall of the sphenoid sinus, whereupon only the sinus mucosa and the dura separate the ICA from the sinus cavity, facilitating rapid spread of infections.[4] Emergence, growth, and rupture of such aneurysms can be rapid, as demonstrated in the present case by the absence of aneurysms on initial imaging after bleeding onset and the subsequent development of a relatively large aneurysm revealed on follow-up CTA just 2 weeks later. In addition, delayed filling and stagnation of contrast agent on DSA are features of pseudoaneurysm[5]

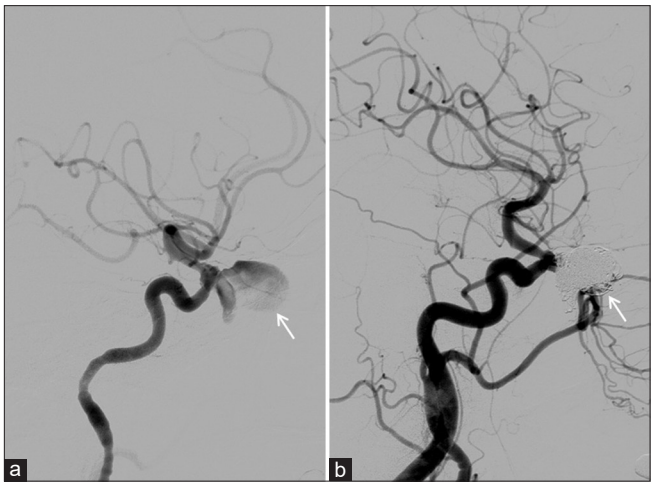

Figure 2: (a) Preoperative digital subtraction angiography (DSA) (lateral view) revealed the large left paraophthalmic internal carotid artery infectious aneurysm (white arrow). (b) Postembolization digital subtraction angiography demonstrated complete exclusion of the pseudoaneurysm (white arrow).

illustrated also in our case [Figure 2]. Most common predisposing condition for infectious aneurysms is valvular heart disease leading to endocarditis followed by meningitis, major surgeries, immunosuppression, dental infections, or IV drugs. Infectious pseudoaneurysms are caused by bacteria, tuberculous bacilli, or fungi.[5] To the best of our knowledge, the rare combination of multiple drug-resistant pathogenic agents such as *P. aeruginosa* and *P. fluorescens* and *A. baumannii* has not been previously reported in the literature and made less effective the role of antibiotic therapy in our immunosuppressed patient. Therefore, selective coiling was feasible in our case due to narrow neck of the aneurysm and has been selected as a lifesaving procedure.

Prompt securing of the aneurysm should be prioritized; we suggest that endovascular occlusion by coiling is a safe and efficient minimally invasive technique of preventing rebleeding, without always necessitating either parent vessel sacrifice or stenting. Stenting may be pursued at a later stage, when hemorrhagic complications are deemed acceptably reduced, if aneurysm recurrence is documented at follow-up. Furthermore, a high index of clinical suspicion is paramount to early recognition. Clinicians should have a low threshold for performing a CTA in patients with persistent epistaxis, as it is rapid and sensitive enough for timely detection of ruptured infectious pseudoaneurysms.

CONCLUSION

Ruptured infectious intracranial ICA aneurysm as a result of infection spread from a paranasal sinus infection is a rare but potentially fatal cause of epistaxis, especially when accompanied by an uncommon anatomic variation in the form of bony dehiscence of the superolateral wall of the sphenoid sinus. Embolization is a minimally invasive technique which can provide efficient treatment in ruptured infectious aneurysms protruding inside the sphenoid sinus and – barring particularities in the morphology and location of the lesion – should be offered as a first-line lifesaving procedure.

Declaration of patient consent

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

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Conflicts of interest

There are no conflicts of interest.

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