

Recurrent Epistaxis of Unknown Origin: the Role of Imaging

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Abstract

We report two cases of patients who presented to the Emergency Room (ER) with a history of recurrent epistaxis. Conservative treatment was provided to both patients and the most common causes of bleeding were excluded. Patients underwent radiological examinations that revealed the presence of an Intracavernous Carotid Artery Aneurysm (ICAA) extending into the sphenoid sinus through the erosion of postero-lateral bony wall. Aneurysms rupture caused massive nasal bleeding. The purpose of our case report is to illustrate as a very common symptom like epistaxis, in a small number of cases can be underestimated: rare and severe causes of nasal bleeding, as ICAA, should therefore always be considered in the differential diagnosis.

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INTRODUCTION

Intracavernous Carotid Artery Aneurysm (ICAA) can be asymptomatic and incidentally diagnosed or it may occasionally cause symptoms and signs of a space-occupying lesion⁽¹⁾. Symptoms include impairment of related cranial nerves (ocular motor nerves, first and second divisions of the trigeminal nerve) due to direct pressure effect^(2,3), Horner's syndrome, various degrees of pain, hearing loss due to hemotympanum⁽¹⁾ and epistaxis due to aneurysm rupture. Epistaxis is a common symptom rarely requiring medical and surgical treatment in ER, often caused by minor and local disease. Nevertheless, in a small number of cases, it can be underestimated: rare and severe causes of nasal bleeding, as ICAA, should therefore always be considered in the differential diagnosis.

Here we report two cases of patients presenting with

recurrent epistaxis caused by ICAA.

CLINICAL PRESENTATION

Case 1

An 88-year-old male, with history of mild hypertension since age of 65, under treatment with anti-platelet aggregation drugs, presented to the ER for a recurrent and severe right-sided epistaxis resulting in temporary loss of consciousness. No history of recent trauma was reported. The remote pathological history revealed two remote transient ischemic attacks at the age of 66 and 72 years. The remainder of his physical examination was unremarkable.

Laboratory analysis pointed out reduction of haemoglobin values (Hb from initial value of 10 g/dl decreased to 7 g/dl; Hct level of 30%) and normal clotting

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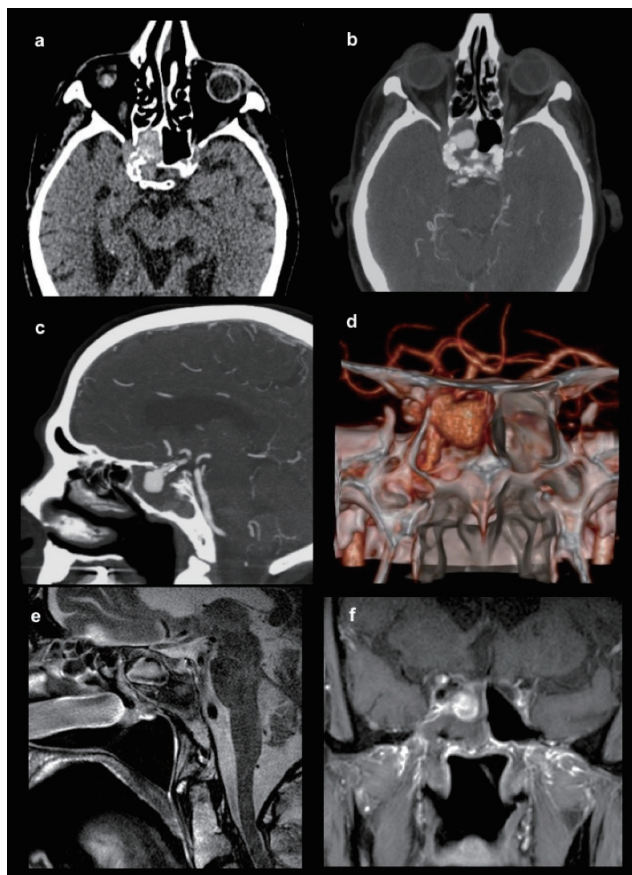
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profile. In a clinical suspect of secondary anemia a blood transfusion was administered.

Due to non-resolution of clinical symptoms a non-contrast Computed Tomography (NCCT) was performed. NCCT revealed heterogeneous hyperdense mass filling the right sphenoid sinus (Figure 1.a). The examination also revealed bone erosion with interruption of sphenoidal posterolateral wall and presence of clot into the nasal cavity. A subsequent CT Angiography (CTA) revealed an ICAA expanding into the sphenoid sinus through the interrupted posterolateral wall (Figure 1.b,c,d). At Magnetic Resonance (MR) imaging, aneurysmal cavity presented heterogeneous signal on T2-Weighted Images (T2-WI) (Figure 1.e) due to partial thrombosis and intra-aneurysmal flow artifacts. Aneurysmal sac was well delineated from surrounding secretions and mucosal inflammatory thickening. Postcontrast T1-Weighted Images (T1-WI) (Figure 1.f) confirmed the presence of an ICAA with a mean diameter of 15 mm, extended into the sphenoidal sinus.



Patient was treated conservatively, due to the high surgical risk. Three days later, he developed a mild left hemiparesis with dysarthria (NIHSS 3): a new CTA revealed a massive spontaneous thrombosis of the aneurysms of Internal Carotid Artery (ICA), which is one of the possible rare complications described in literature⁽⁴⁾.

Case 2

87-year-old male, without history of severe head trauma and with a recent episode of epistaxis that required front and back nasal padding, was admitted to ER because of massive nasal bleeding. Patient did not refer coagulation pathologies or treatment with anti-platelet aggregation drugs. A NCCT scan revealed the presence of a mildly hyperdense lesion with a mean diameter of 1.8 cm, in the left cavernous sinus, protruding into the sphenoidal sinus cavity (Figure 2.a). The bone window disclosed discontinuity of superior and lateral walls of sphenoidal sinus (Figure 2.b). At the MR, the lesion showed signal partially hyperintense on T1-WI (Figure 2.c) and hypointense on T2-WI, mimicking a fungal or

Figure 1. a. NCCT scan shows a heterogeneously hyperdense mass filling the right sphenoid sinus cavity
 b, c. CTA axial and sagittal MIP images clearly depict the intracavernous aneurysm expanding into the sphenoid sinus through the deiscent posterolateral wall
 d. 3D CTA image (anterior view) nicely depicts location and orientation of the aneurysm
 e. Aneurysmal cavity exhibit heterogeneous signal on sagittal T2-WI, due to partial thrombosis and internal flow artifacts. The lesion is well demarcated from the surrounding inspissated secretions.
 f. Coronal postcontrast T1-WI. The use of contrast significantly improves the identification of the aneurysmal sac.

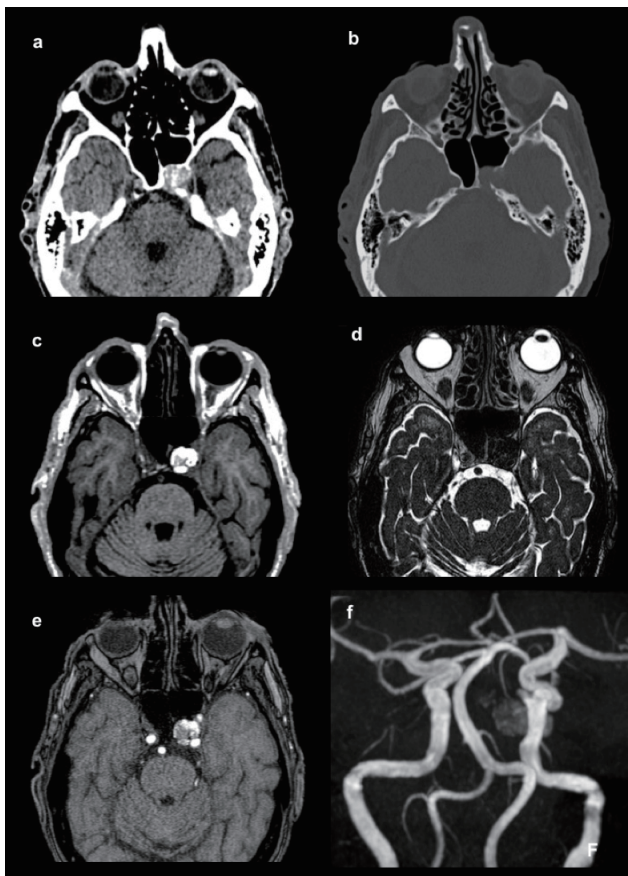


Figure 2. a. NCCT scan reveals a mildly hyperdense lesion protruding into the left sinus cavity.
 b. Bone window well demonstrates the erosion of sinus wall caused by the long-standing aneurysm.
 c. Axial T1-WI highlights the bright signal of methemoglobin within the aneurysmal thrombus.
 d. Hypointensity of the aneurysm on T2-WI may mimic a fungal or neoplastic disease.
 e. 3D TOF MRA source image nicely shows the small communicating part of the aneurysm, brighter than the large thrombotic component.
 f. 3D TOF MRA volume rendered image visualizes the aneurysm due to the pseudoenhancement effect of methemoglobin within the thrombus

neoplastic disease (Figure 2.d). MR Angiography Time Of Flight (TOF MRA) sequence (Figure 2.e,f) demonstrated ICAA with an extensive parietal thrombosed component, characterized by bright signal due to the paramagnetic effect of methemoglobin.

DISCUSSION

From an anatomopathological point of view, a distinction must be made between true and false aneurysms or pseudoaneurysms, because, although they may seem similar at imaging, pseudoaneurysm is a blood harvesting without vascular wall, contained by the perivascular soft tissues or by residual adventitia. Furthermore, pseudoaneurysms are mostly caused by trauma, inflammation, radiation therapy or skull base surgery [5]. Otherwise true aneurysms are bound by all three layers of the vessel wall (intima, media and adventitia) and the risk of rupture is proportional to their size. In both our

cases the patients have no history of head trauma and the cause of bleeding was identified as true aneurysms of the intracavernous tract of the internal carotid artery, unlike most cases in literature describe pseudoaneurysms due to trauma on this site.

True aneurysms are most often secondary to atherosclerosis and are classified by their gross appearance in Saccular or ‘Berry’ aneurysms (SAs) and Fusiform aneurysms (FAs). SAs are the most common types and typically arise eccentrically at vessel branch points, while FAs are focal dilatations that involve the entire circumference of the vessel, extend for relatively limited distances.

Intracranial true aneurysms are relatively common, with a prevalence of approximately 4%. They are located in 30% of cases on Anterior Communicating Artery (ACoA), in 25% on Posterior Communicating Artery (PCoA) and in 20% on Middle Cerebral Artery (MCA). Most aneurysms are asymptomatic and have a good

prognosis, but they can grow unpredictably; moreover, even small aneurysms carry a risk of rupture. Unruptured aneurysms may cause symptoms, mainly due to mass effect towards adjacent structures. However, the most severe complication is related to their rupture, leading to a subarachnoid hemorrhage.

ICAA are rare, accounting for 3 to 5% of all intracranial aneurysms and 14% of all aneurysms arising from the ICA. The majority of ICAA are less than 2.5 cm in diameter and rarely cause epistaxis [6]. However, this symptom is characteristically present when ICAA is located posteriorly to the wall of the sphenoid sinus, often less than 2 mm thick. As in our reported cases, a long-standing aneurysm can easily erode the bone, invade sphenoid sinus and cause epistaxis⁽⁷⁾. The tamponade effect of the clot within the sinus can arrest the initial hemorrhage, but the subsequent lysis of the hematoma results in recurrent and often catastrophic bleeding⁽⁸⁾.

Clinically, epistaxis is a common otolaryngologic cause of hospital admission: during life, almost 60% of adults experience an epistaxis episode and 10% or fewer seek medical attention. Only a small percentage of them need surgical intervention. Massive epistaxis due to non-traumatic ICAA is extremely uncommon and potentially fatal, with a restricted number of cases reported in the literature⁽⁹⁾. When epistaxis is the main presenting symptom, the diagnosis may be delayed or overlooked due to the common nature of this symptom. Awareness of this potential condition, in patients with massive epistaxis is important to direct the clinician to perform radiological investigations in order to make the diagnosis and carry out effective treatment.

Radiological diagnosis is not always simple; in fact, ICAA can mimic an aggressive inflammatory, as fungal infection particularly, or neoplastic masses arising from sphenoid sinus, both potentially appearing as a high attenuating and locally destructive masses⁽⁵⁾.

Even in our case, NCCT didn't help in differentiating between inflammatory or vascular origin of the tissue.

MR imaging can aid in distinguishing vascular structures from surrounding tissues. Both in Fast Spin-Echo T2-WI and Gradient-Echo T1-WI sequences vessels are well depicted, showing the characteristic dark (flow void artifact) or very bright signal, respectively. TOF MRA, provides optimal assessment of vascular lesions:

source images and post processed data (Maximum Intensity Projection, Multi Planar Reconstruction, and Volume Rendering; MIP, MPR and VR) enable, as for CTA, accurate evaluation of cerebral vessels.

CTA is a less invasive and alternative technique to conventional catheter angiography; it is usually performed as the initial investigation, although the latter represents the gold standard to determine the feasibility of endovascular therapy⁽¹⁰⁾. Various endovascular therapeutic options, including balloon occlusion and stent-assisted coiling, have been described with favorable technical outcomes⁽⁹⁾.

CONCLUSIONS

Severe epistaxis due to non-traumatic ICAA is extremely uncommon and potentially fatal. In cases with no history of head trauma, the diagnosis may be often elusive and delayed by the common nature of this symptom. When recurrent epistaxis is accompanied by history of loss of consciousness, impairment of cranial nerves, conductive hearing loss, without any systemic cause or local Ear, Nose and Throat (ENT) pathology, ICAA should be included in the differential diagnosis. Its management requires a multidisciplinary approach, being the mortality up to 30%⁽²⁾. Clinicians should be aware about the crucial role of imaging. Differential diagnosis is challenging: ICAA can be mistaken with inflammatory or neoplastic diseases of the sphenoid sinus⁽⁵⁾, thus its correct recognition is mandatory to avoid inopportune and dangerous diagnostic procedures (i.e. biopsy sampling) and severe consequences.

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