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Case report

Iatrogenic bleeding of extracranial internal carotid artery aneurysm mimicking peritonsillar abscess

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Abstract. *Background and aim*: Extracranial Internal Carotid Artery Aneurysms are very rare. They can manifest with variable and unspecific clinical instances including neurologic symptoms associated with pulsatile cervical mass as the most frequent ones. This way, in presence of a mass in the oropharynx or neck, an aneurysm of the Internal Carotid Artery is not often considered as a prior diagnosis, therefore some of them are only discovered during surgical procedures. Here we describe a case of Extracranial Internal Carotid Artery Aneurysm misdiagnosed as a Peritonsillar Abscess which underwent surgical drainage. The purpose of this article is to emphasize factors which should be considered in differential diagnosis before doing inappropriate operative procedures. We will also focus on the effectiveness of Angiography, angio-CT and angio-MR in diagnosing the aneurysm and which factors to consider when choosing the best surgical or endovascular treatment. *Method:* Retrospective case report. *Results:* An Extracranial Internal Carotid Artery Aneurysm was misdiagnosed as a Peritonsillar Abscess which underwent surgical drainage with consequent profuse haemorrhage. An emergency tracheostomy was immediately performed. The patient was moved to the Interventional Neuroradiology Department where angiography revealed a double kinking and a giant aneurism of the Internal Carotid Artery. Endovascular permanent occlusion of the Internal carotid Artery was performed with detachable coils. The patient reported neither neurologic sequelae nor haemorrhage recurrence during the 2–year follow-up. *Conclusions:* Aneurysmatic dilation of Extracranial Internal Carotid Artery in the parapharyngeal space may determine, especially in presence of kinking, a medialization of the lateral oropharyngeal wall, mimicking a PTA. A high index of suspicion is mandatory for diagnosis. (www.actabiomedica.it)

Key words: Internal Carotid Aneurysm, Internal Carotid Kinking, Peritonsillar Abscess, Endovascular Treatment

Introduction

Extracranial Internal Carotid Artery Aneurysms (EICAA) are characterized by a local increase in vessel diameter >50% when compared to the standard values for appropriate vessel measures (1). EICAA are rare arterial lesions with incidence about 0.8% to 1% of all arterial aneurysms, more frequent in males with a 2-3:1 rate (2).

Atherosclerosis is the leading cause and other aetiologies include fibromuscular dysplasia, trauma, previous carotid surgery, vasculitis and infections.

Classification of EICAA have been reported by Attigah et al. (3) and by Chen et al (4), who proposed the so called PUMCH classification (Peking Union Medical College Hospital). Both classifications are helpful in the choice of the type of treatment: surgical or endovascular.

Clinical manifestations include neurologic disorders, symptoms related to the compression of local structures and haemorrhage. Neurologic disorders are the most common ones and they typically include nerves palsies, cranial nerve dysfunction, transient ischemic attacks or complete strokes (1) (5). Differential diagnoses include neoplastic aetiologies, such as oropharyngeal and parapharyngeal space tumours, and infections, the latter represented in particular by phlegmon and Peritonsillar Abscess (PTA). Duplex Doppler Ultrasonography is the first line diagnostic method of choice in suspected EICAA. Angio-CT, angio-MRI and digital subtraction angiography are used for a more detailed pre-operative evaluation. Treatment of EICAA has progressively changed during the years, moving from surgical to endovascular treatment (1)(5).

In this article we present a case of an EICAA misdiagnosed as a PTA, which underwent surgical drainage and caused profuse bleeding. The patient has been treated by endovascular occlusion of the internal carotid artery (ICA) with good results.

Case Report

An 80-year-old woman who had had dysphagia, sore throat and earache for a few days was referred to our centre for a suspected PTA. The patient had been previously assessed by the primary care physician and treated with anti-inflammatory drugs and antibiotics without any benefit. Anamnesis was negative for trauma or previous vascular surgery in the cervicofacial region. Hypertension was reported as the only chronic disease on medication.

Routine blood tests were made by the triage team as usual, with fundamentally normal results for the tested parameters, including acute phase reactants and white blood cells.

The clinical examination showed a medial displacement of the right palatine tonsil with pharyngeal mucosa hyperaemic and floating on palpation. There was not evident spontaneous pulse in the pharynx and in the neck. The oral opening was reduced by a trismus but the uvula size was normal and fever was absent.

Considering this clinical presentation, the otolaryngologist confirmed the diagnosis of PTA and proceeded to perform a surgical drainage of the peritonsillar space under local anaesthesia. An endoral and profuse parapharyngeal haemorrhage occurred immediately at the incision. An emergency tracheostomy was at once successfully performed to ensure ventilation and protect the airways from inhalation of blood. In suspicion of a iatrogenic injury of the carotid artery, the patient was moved to the Interventional Neuroradiology Department. A preliminary evaluation with angio-CT could have been useful but, due to the emergent clinical conditions, the patient was immediately transferred to the angio-room. Angiography was performed under general anaesthesia through a transfemoral approach: the cervical segment of the ICA appeared tortuous and elongated, with a double kinking and a giant aneurysm measuring 5 cm in diameter (Fig. 1a,b).

The vessel downstream the aneurysm was poorly opacified and the flow was very slow. We classified the aneurysm as Attigah type I and PUMCH type II/b. A functional evaluation of the circle of Wills was done through manual compression of the right common carotid artery and subsequent injection, first into the left dominant vertebral artery (Fig. 2a) and then into the contralateral left ICA. Both the posterior and anterior communicating arteries were patent. The whole right ICA vascular territory was compensated by the vertebrobasilar system, whereas only the right anterior cerebral artery vascular territory was compensated by the contralateral ICA. All the angiographic phases were symmetric, without any delay in the appearance of the venous phase on the site of the occlusion in comparison to the vascular territory of the afferent vessel (Fig. 2b).

Due to the high position of the aneurysm (which was located above the line of Blasedale) (4) and the advanced age of the patient, we decided for the least invasive form of endovascular treatment. We chose to perform permanent occlusion of the ICA in presence of the good angiographic tolerance test, based on the symmetry of the venous phase (6). Another factor which led us to choose a permanent occlusion of the CIA was the impossibility to insert a covered stent, due to the presence of kinking in the cervical segment of the ICA proximal to the aneurysm.

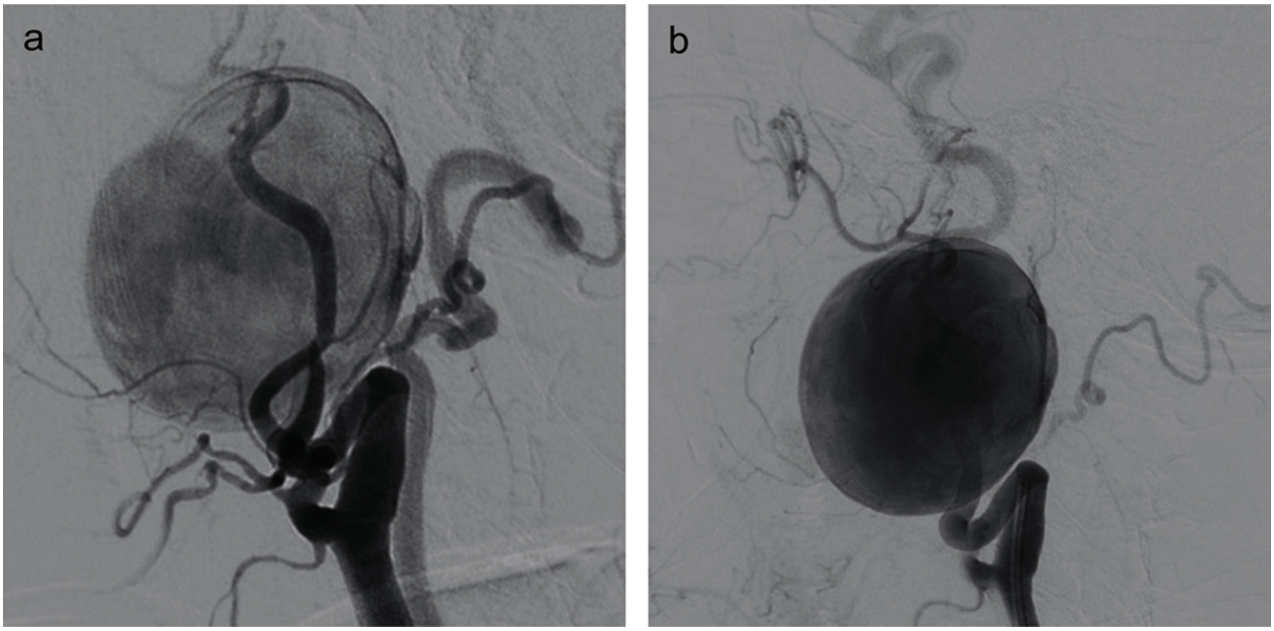

Figure 1. Injection of the common carotid artery, lateral projection. a) early angiographic phase showing kinking of the internal carotid artery proximal to the aneurysmal sac which fills very slowly; the internal carotid artery downstream is not yet opacified; visualization of the homolateral vertebral artery by reflux. b) late angiographic phase showing the aneurysmal sac completely opacified and downstream visualization of the internal carotid artery.

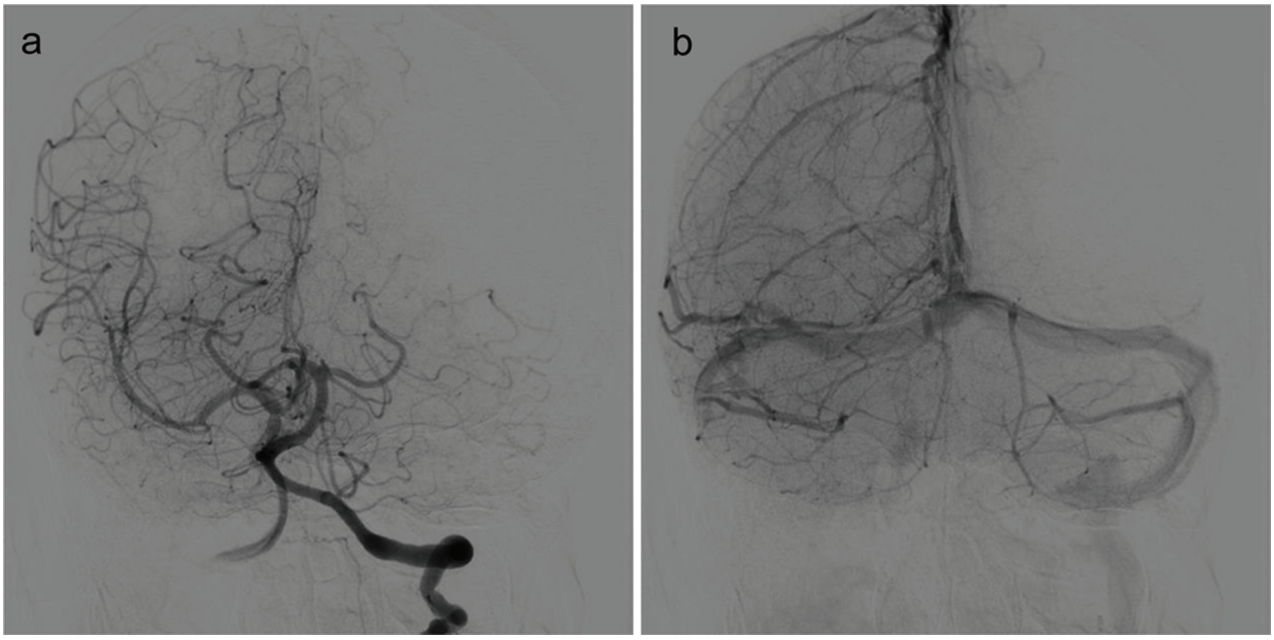

Figure 2. Injection of the left vertebral artery in the anteroposterior view after carotid occlusion. a) arterial phase showing rapid filling of the vascular territory of the right internal carotid artery which is synchronous to the filling of the homolateral posterior cerebral artery. b) venous phase showing symmetric filling of the veins of the vertebrobasilar territory and the veins of the right internal carotid territory without any delay.

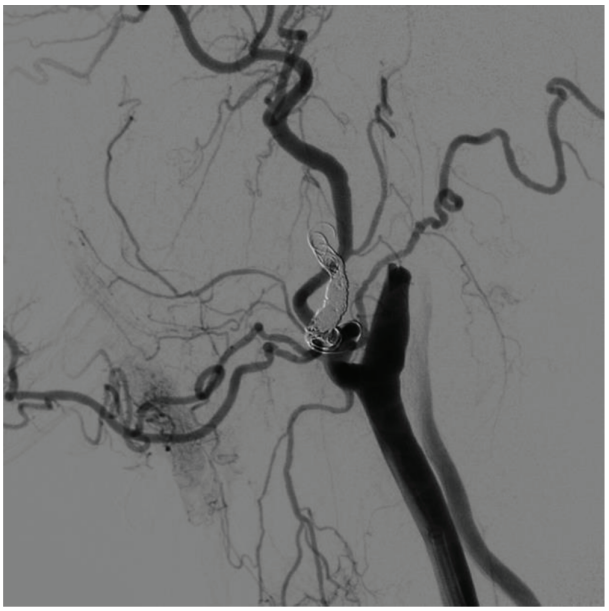

Figure 3. Injection of the right common carotid artery (lateral view) after coil occlusion of the internal carotid artery, proximal to the aneurysm.

A 6 French guiding catheter was inserted into the ICA and occlusion of the parent vessel proximal to the aneurysm was performed with detachable coils, through a microcatheter (Fig. 3).

Although advisable, a trapping procedure (occlusion of the parent vessel below and above the aneurysm) was considered too difficult because of the impossibility to reach the normal segment of the parent vessel downstream the aneurysm.

Subsequently, the patient was strictly monitored in the Intensive Care Unit Department for a few days in order to avoid dehydration and hypotension. Finally, she was transferred back to the Department of Otolaryngology for swallowing and airways rehabilitation. At last she was decannulated and discharged from the hospital. The post-procedural angio–CT performed on the following day showed complete exclusion of the aneurysm and regular opacification of the intracranial circulation. The patient reported neither neurologic sequelae nor haemorrhage recurrence during the 2 –year follow-up.

Discussion

In presence of a mass in the oropharynx or neck, the possibility of an EICAA is not often considered as prior and some aneurysms are discovered during surgical procedures (7). Differential diagnosis includes abscess (PTA), an inflammatory or neoplastic lesion of a tonsil, laryngopathy or even a neck tumour. Szopinski et al. reported a similar case of a patient with haemorrhage after an attempt of drainage of suspected PTA (7); Karov, instead, a case of spontaneous rupture of EICAA previously diagnosed as a PTA and, afterwards, as a carcinoma of the pharynx with metastasis (8).

EICAA can present with multiple but unspecific symptoms including dysphagia, headache, trismus, odynophagia and hoarseness (9). Pulsation of the neck may be totally absent, like in our case, or at least weakened (7).

Most patients suspected for PTA have already been treated with systemic antibiotics before being admitted (10). Therefore, the presence of low levels of acute phase reactants or the absence of uvula oedema could mislead the diagnosis if we consider them as a medical therapy consequence of PTA rather than an absence of infective pathology.

In presence of a mass in the oropharynx or neck the existence of risk factors for vascular pathologies should be considered. In our case, age and hypertension were present, but also the patient's history of previous trauma or vascular surgery, neck irradiation, infections, fibromuscular dysplasia or vasculitis should be investigated. Neurologic evaluation should also exclude evidence of embolic stroke and cranial nerve deficits which are frequently counted among first presentations of aneurysms (7). In a retrospective analysis, Radak et al. revealed that patients with ICA kinking had aneurysms of a greater diameter compared to patients without kinking (5). This phenomenon is probably related to the more medial and deeper course that the kinked Carotid Artery takes, which can delay the aneurysm diagnosis.

Most of the authors agree that Duplex Doppler Ultrasonography is the simplest and most non-invasive diagnostic method in suspected EICAA. Therefore, it should be the first one to be used despite its low sensitivity for small aneurysms located in the distal ICA at the base of the skull. Although angio-CT and angio-MR can also accurately assess ICA aneurysms (11), arteriography remains the gold standard to show the anatomy of the ICA and the collateral flow through the circle of Willis (7).

For many years, the first line of treatment of EICAA has been open aneurysmectomy with end-to-end anastomosis or graft interposition (1). In recent years though, endovascular approaches have also been successfully used in case of high risk of nerve damage or in case of aneurysms extending into the skull base, above the line of Blasedell (11). The choice of a particular endovascular procedure is determined by the modality of clinical presentation of the aneurysm, ruptured or unruptured, by the anatomy of the ICA and by the results of the angiographic tolerance test to parent vessel occlusion. Various kinds of endovascular treatment have been proposed: bare stent insertion with or without coil embolization, intraluminal flow diversion, permanent occlusion of the ICA with balloons, coils or plugs and insertion of a covered stent. In our case, based on the assessment of the venous phase delay, we decided for permanent occlusion of the ICA with coils. What led us to this choice was the emergent clinical situation which presented difficulties to navigate covered stents into the kinked ICA and the good tolerance to permanent ICA occlusion (6)(12).

In conclusion, aneurysmatic dilation of ICA in the parapharyngeal space may determine, especially in presence of kinking, a medialization of the lateral oropharyngeal wall, mimicking a PTA. Presence of typical PTA-associated symptoms like pain and trismus, lack of pulsation or neurologic symptoms, should not encourage the otolaryngologist to exclude a diagnosis of EICAA. Nevertheless, a high index of suspicion is always mandatory for diagnosis.

Conflict of Interest: Each author declares that he or she has no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article.

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