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Case Report

Refractory Haematemesis Resulting from ICA Mycotic Pseudoaneurysm: Managed by Endovascular Coiling

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Abstract: Pseudoaneurysms of extra cranial carotid artery are rare and are most commonly associated with blunt or penetrating trauma. Other cause includes iatrogenic origin, inflammation, infection, vasculitis, tumor, and arteriosclerosis. Pseudoaneurysm of ICA at the extracranial segment is a rare complication of neck infections. The treatment of carotid artery mycotic pseudoaneurysm is complex. The typical management of an mycotic pseudoaneurysm is twofold: systemic antibiotic therapy and surgery, with either a traditional by-pass or a ligation of ICA. Endovascular therapy of a non-infected and infected carotid artery pseudoaneurysm has been increasingly used, with this treatment the ICA lumen may be better preserved. We report a case of 11 yr old male who presented with massive haematemesis from mycotic pseudoaneurysm secondary to tubercular cervical adenitis managed successfully by endovascular coiling. **Keywords:** Internal carotid artery, endovascular therapy, mycotic, pseudoaneurysm

INTRODUCTION

The word aneurysm is derived from Latin word *aneurysma*, meaning dilatation. An aneurysm is an abnormal widening or ballooning of a portion of an artery due to weakness in the wall of the blood vessel. A true aneurysm is one that involves all three layers of the wall of an artery (media, intima, adventitia). A pseudoaneurysm, or false aneurysm, occurs as the result of a leaking hole in a blood vessel. A hematoma forms outside the arterial wall such that leaking blood is contained by the surrounding tissues while remaining in continuity with the breached arterial lumen.Extracranial internal Carotid Artery aneurysms are defined as localized increases of the caliber of more than 50% as compared with the reference values of the ICA (0.55 \pm 0.06 cm in men; 0.49 \pm 0.07 in women) [1].

Pseudoaneurysms (PAs) comprise only 14% of extra cranial carotid artery aneurysms in the reviewed literature [2]. PAs are mainly secondary to previous end arterectomy, other contributing factors of Pas may be trauma, iatrogenic central venous cannulation and infections. True aneurysms are most commonly due to atherosclerosis or fibromuscular dysplasia. Other rare contributing factors to aneurysm formation include neck irradiation, neurofibromatosis, Marfan's Syndrome, Bechet's Syndrome and Takayasus's arteritis. 30% to 60% of extra cranial aneurysms are symptomatic for thrombo-embolic focal or non focal symptoms; more rarely cranial nerve compression may result in neurological deficits. Other adverse events may be rupture, with massive hemorrhage [3]. Here we report a case of refractory haematemesis in a young boy diagnosed to have a mycotic ICA pseudoaneurysm managed successfully by endovascular coiling.

CASE REPORT

A 11year old boy presented with massive haematemsis (bright red) in emergency department of our hospital. He had history of fever since last 10days.On admission his BP was less than 60mm of Hg with feeble pulse. On clinical examination there was a palpable, pulsatile left sided swelling in neck in addition multiple,palpable,non-tender,discrete there were cervical lymph nodes. Patient was resuscitated & stabilized. Pathological investigation revealed leucocytosis, relative lymphocytosis, raised ESR and anaemia. Imaging workup included duplex sonography followed by a cerebral and neck computed tomography. Doppler revealed a large, saccular out pouching from left distal ICA with swirling of blood flow within (Yin Yang phenomenon). Pulse Doppler shows to-and-fro waveforms. CECT Neck revealed a large, lobulated pseudoaneurysm (PA) in the left carotid space originating from the left ICA with irregular indistinct margins & peri-aneurysmal oedema (Fig 1). The pseudoaneurysm was associated with multiple perianeurysmal lymph nodes (Fig 2a), few of them were caseating. There was associated perianeurysmal gas (Fig 2b). Angio-CT scan revealed a narrow neck, saccular, "diverticulum-like" aneurysm from distal ICA, developing from the side of the carotid wall (Fig 3, 4). Evaluation of the collateral pathways in the circle of Willis was done. Endovascular management of pseudoaneurysm was planned. Our choice of the endovascular treatment was chiefly influenced by the unfavorably deep location of the PA near the skull base thus making conventional surgery very risky and critical patient condition. Dual antiplatelet therapy was started with clopidogrel and aspirin.

On DSA the pseudo-aneurysm was confirmed arising from medial wall of left distal extra-cranial ICA (Fig 5) in addition there was significant stenosis, measuring approximately 91%, just distal to the pseudoaneurysm. On basis of DSA findings endovascular parent artery occlusion by positioning coils distally and proximal to pseudoaneurysm was decided.

The first coil deployed was a standard 4 x 10mm coil in distal ICA.Additional coil of diminishing size was then placed in distal ICA, close to the pseudoaneurysm. A selective microcatheter injection of the left internal carotid artery after coiling shows the pseudoaneurysm was still filling up & was not completely obliterated. The last three 4 x 10 mm coils were detached and placed without difficulty in proximal ICA segment. An unsubstracted left internal carotid angiogram after coiling shows complete obliteration of the left ICA and non-filling of pseudoaneurysm sac (Fig 6).

Post endovascular coiling angiograms of the head and neck was then obtained, which documented complete occlusion of left ICA with good opacification of left PCA through basilar artery and left MCA through contralateral collateral circulation (Fig 7).

The patient did well after the procedure, with no new neurologic deficit. His heparin anticoagulation was continued overnight and was stopped the following morning. He returned home the day after his procedure, receiving dual antiplatelet therapy and anti-tubercular therapy. Six months after the endovascular procedure this young boy was doing well, attending school, and participating in sport activities.

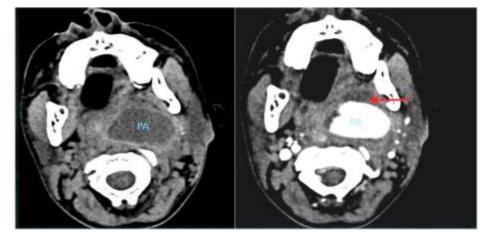


Fig.1(a & b): MDCT axial images(a) plain (b) contrast reveals a large, lobulated, pseudoaneurysm in the left carotid space space originating from the left ICA with irregular margins & peri-aneurysmal edema (arrow)

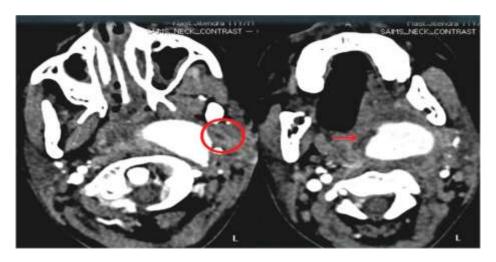


Fig. 2(a & b): CECT Axial images-The pseudoaneurysm was associated with multiple perianeurysmal lymph nodes (O), few of them were caseating. There was associated perianeurysmal gas (arrow).

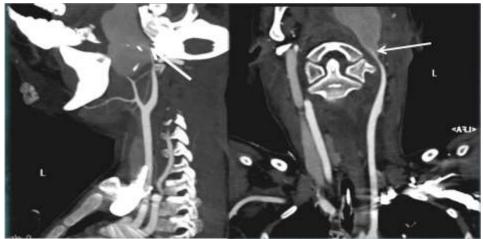


Fig. 3(a & b): CT Angiographic images demonstrating a large pseudoaneurysm (arrows) from distal left ICA



Fig.4 (a,b & c): CT Angiography VRT Images demonstrating a narrow neck,saccular, pseudoaneurysm from left ICA



Fig. 5: DSA demonstrating a large pseudoaneurysm from left ICA with significant stenosis (91%) of parent vessel

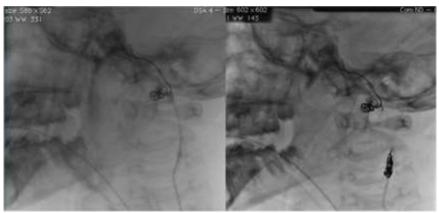


Fig. 6(a & b): Unsubstracted angiography images demonstrating endovascular coil in distal segment and proximal segment of left ICA with its complete occlusion and non-filling of pseudoaneurysm



Fig.7 (a-d): Post coiling CT angiography images documenting complete occlusion of left ICA (a & b). Good opacification of left PCA (c) and left MCA (d) through basilar artery and contra lateral collaterals respectively

DISCUSSION

The causes of a pseudoaneurysm are varied but are most commonly associated with blunt or penetrating trauma. Other cause includes iatrogenic origin, inflammation, infection, vasculitis, tumor, and arteriosclerosis; however, there are also cases of unknown origin. The common causes of vasculitis resulting in a pseudoaneurysm include Takayusu's disease, polyarteritis nodosa, Kawasaki disease, and Behcet's disease [4].

PA of ICA at the extra cranial segment is a rare complication of deep neck infections. Compared to a

true aneurysm, the PA has no complete native arterial wall. PA is composed of extra-vasated blood that leaked from the area of vessel erosion and is surrounded by inflammatory and fibrous tissues. PA of ICA in children is more frequent. Children are more susceptible to arteritis [5]. Infection can reach the wall of the carotid artery following a peritonsillar abscess or pharyngitis.

Another pathway for infection may be by septicemia with invasion of the vasa vasorum. In our patient, the PA was a result of the tubercular cervical adenopathy reaching the left ICA adventitia. Ischemia of the carotid artery wall led to its rupture and development of PA. A mycotic carotid PA most commonly presents as a growing, pulsatile cervical mass, dysphagia, odynophagia, and fever. Less frequently, lower cranial nerve palsies, Horner's syndrome or trismus may occur. Severe and life-threatening complications may include a artery rupture, intermittent carotid massive nasopharyngeal hemorrhage, and septic or non-septic embolic events leading to a neurological deficit. The usual interval between the infection and the PA development is between 2 to 8 weeks. Imaging characteristics of mycotic aneurysm include large, lobulated pulsatile neck mass, irregular indistinct margin of mass, pediatric age group, perianeurysmal odema, perianeurysmal & cervical lymphadenitis and perianeurysmal gas [6].

The treatment of carotid artery PA is complex. The typical management of an infected PA is twofold: systemic antibiotic therapy and surgery, with either a traditional by-pass or a ligation of ICA [7-10]. Endovascular therapy of a non-infected and infected carotid artery PA has been increasingly used [8, 11, 12]. With this treatment the ICA lumen may be better preserved. Several approaches are available. The novel technique [13] was a parent artery occlusion achieved by positioning detachable balloons distally and proximally to the lesion. However, this approach demands preliminary evaluation of the collateral pathways in the circle of Willis. The occlusion test requires a fully conscious patient to monitor neurological deficits. The inherent risk of the occlusion test includes development of neurological deficits and or failure to identify a delayed ischemia. Another endovascular approach preserving the carotid artery lumen is a stent or stent-graft implantation with or without a coil deposition to the PA. Since the PA lacks a true arterial wall, a potential risk of compaction and dislocation of the coils is always present. A simple stent or a stent-graft implantation is regarded to be more effective and faster treatment [6, 9, 14, 15].

Our choice of the endovascular treatment was chiefly influenced by the unfavorably deep location of the PA near the skull base thus making conventional surgery very risky and critical patient condition. Moreover, patency of the collateral pathways in the circle of Willis on preliminary examination was reassuring. Because of significant stenosis (91%) of ICA just distal to pseudoaneurysm, parent artery occlusion by positioning coils distally and proximal to pseudoaneurysm was planned and achieved successfully.

CONCLUSION

The possibility of a pseudoaneurysm must be considered when a patient complains of a pulsating neck mass, even if there is no history of trauma, as was the case in this study. Based on our results, endovascular management can be an effective alternative to surgery for the treatment of extra cranial carotid pseudoaneurysms.

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