Carotid artery pseudoaneurysm rupture: A case report

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Case report

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Background
Carotid artery pseudoaneurysms may cause massive oropharyngeal hemorrhage and upper airway obstruction due to mass effect. Life threatening complications may arise such as difficult airway or traumatic rupture of the aneurysm during the course of management. Here, we present a case of a 44-year-old male patient who developed multiple carotid pseudoaneurysms post-radiation therapy due to nasopharyngeal carcinoma. We would like to take this as an opportunity to raise awareness among clinicians on the importance of triaging such patients with a multidisciplinary approach, specifically early involvement of anesthesiologists, radiologists, intensivists, ENT surgeons and vascular surgeons to optimize care and outcome.

Keywords: Coiling, carotid artery, pseudoaneurysms, stenting, post radiation, massive hemorrhage, vascular surgery

Introduction

Pseudo aneurysms (PSA) of internal carotid arteries are rare (1). Before the antibiotic era, middle ear infections and tonsillar infections, tuberculosis, syphilis were the most common cause of carotid artery aneurysms with a significant risk of rupture and associated high mortality rate (2) . In modern day practice, they usually occur as a sequela of trauma, dissection, atherosclerotic degeneration, previous carotid surgery or post radiation therapy for head and neck malignancies. This entity can often be asymptomatic, and discovered incidentally. Currently available treatment options are surgery and endovascular interventions, the latter being less invasive. The main goals of treatment are to prevent PSA rupture, embolic events and procedure related neurological deficits. It is essential to have a timely diagnosis in such cases to prevent life threatening complications and mortality (3). Our case presentation will demonstrate the importance of a multidisciplinary clinical approach in achieving this goal.

Case report

Episode I

We present a 44 year old male patient, known to have nasopharyngeal carcinoma diagnosed 6 months ago for which he received concurrent chemo- and radiotherapy; external beam radiotherapy to the head and neck 70.00gy/ 35 cycles, and weekly cisplatin 40mg/m2 for two months. He came to the emergency department with a two-day history of cough, fever and difficulty in breathing. On examination he had altered mental status, in respiratory distress - with notable stridor. Blood gas analysis suggested a hypercapnic type 2 respiratory failure with CO2 retention of up to 74 mmHg .Preliminary diagnosis was respiratory tract infection with the need to rule out mechanical obstruction of upper airway. A decision was made for emergency intubation and mechanical ventilatory support. After several unsuccessful intubation attempts, massive oropharyngeal hemorrhage and epistaxis were noted. Airway trauma, supraglottic tumor and severe fibrosis with distorted anatomy were thought to be the potential culprits for the new onset bleeding episode. Emergency tracheostomy was performed. Upon completion of the procedure, patient suffered a cardiac arrest due to significant blood loss, estimated around 1.5-2 litres. Cardiopulmonary Resuscitation was commenced as per advanced cardiovascular life support (ACLS) protocols going for 4 cycles each 3 mins apart before achieving return of spontaneous circulation. Patient was admitted to intensive care unit (ICU).

On Day 1 and 2 in ICU, multiple bleeding episodes occurred with significant loss of volume up

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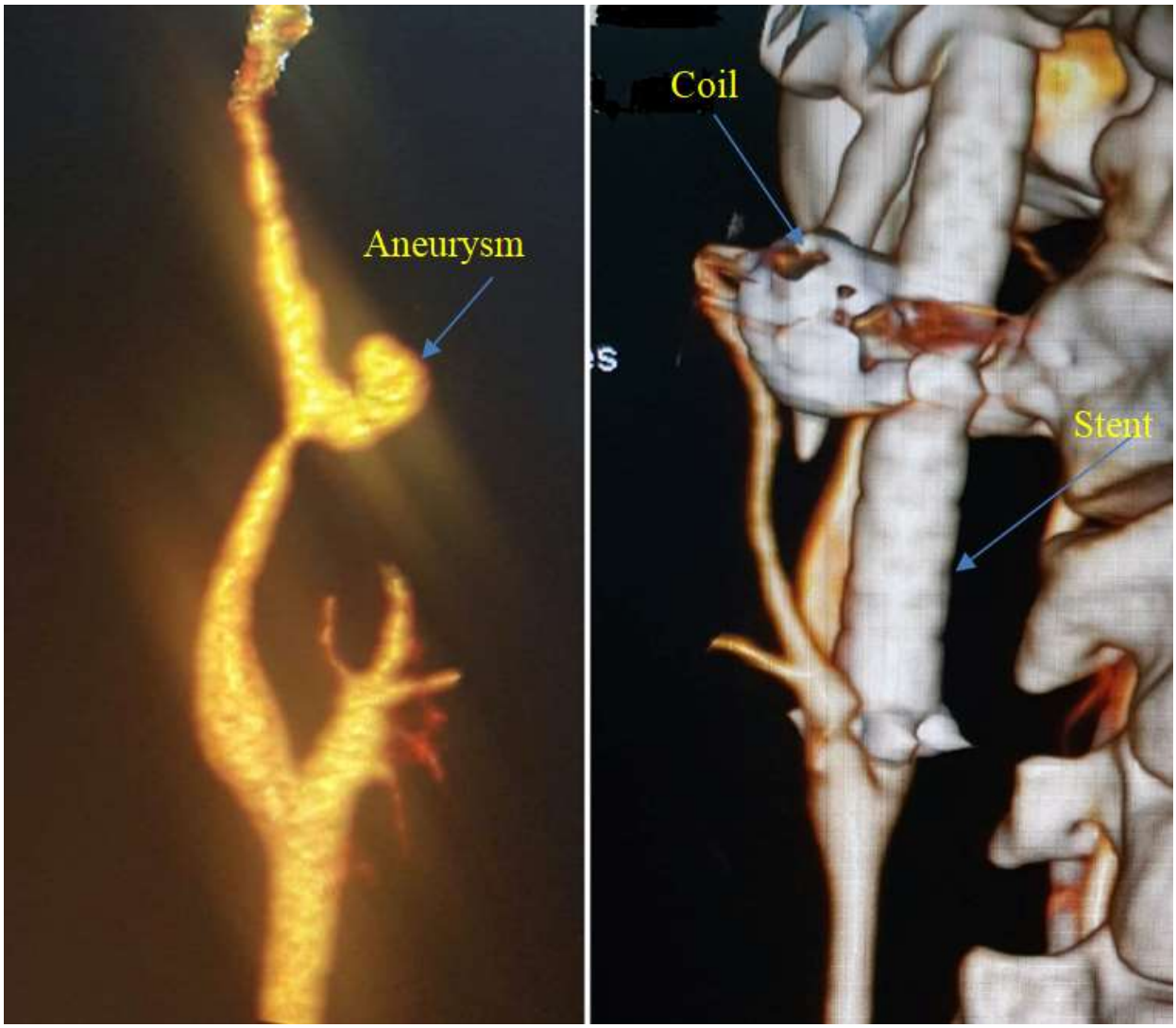

Figure 1. (left). Right Internal carotid artery Aneurysm with Tortuity as seen on CT angiography. (Right) Intraoperative repair with coiling and placement of a covered stent graft.

to a pint (500cc) with each episode. Massive transfusion protocol was activated. Episodes were managed by application of external pressure to the pharyngeal area with the help of adrenaline and tranexamic acid treated gauzes, Magill forceps and Miller 4 laryngoscope blade. CT angiogram of the intra and extra cranial vessels was done that revealed right internal carotid bi-lobed pseudoaneurysm measuring (12.78×5.79mm) large and (5.27×4.31mm) small, facing anteriorly and likely source of bleeding. Figure 1a The intracranial circulation was all intact. Coil embolization was done for one of the lobes of the pseudoaneurysm using 10×30mm and 8×30mm coils. The second lobe was technically difficult to coil and embolize because of a wide neck. Embolization of left internal maxillary artery was also performed by the interventional radiologist in the same angiography session. These interventions temporarily stopped the bleeding while waiting for availability of an appropriate size stent graft. Figure. 1b .Within 12 hours he started having recurrent bleeding episodes once again. A detailed direct laryngoscopy examination was done by an ear-nose and throat surgeon. Significant friability of the tissues was noted and attributed to the post radiation effect. A longitudinal tear was observed at the posterior pharynx and a small mass was found around epiglottis for which biopsy was taken. However, still the source of bleeding could not be identified. Patient was then taken for a CT angiogram which showed right internal carotid artery aneurysm leakage, lesion noted as 17 mm and lobulated. Bleeding episodes still continued as a vascular surgeon was invited to the case. Internal carotid artery stenting was planned and performed successfully. A stent graft 6mm × 80mm (Fluency Bard®) deployed to cover the bilobed pseudoaneurysm. Bleeding episodes stopped. He was started on dual antiplatelet therapy. MRI of the head revealed no mass in the nasopharyngeal area. Brain images showed features of cortical infarcts.

Following his significant clinical improvement, the patient was discharged to the ward. He was independent and mobile with no neurological symptoms.

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Figure 2 (Left). Preoperative images of the right common carotid aneurysms. (Right). Post-operative images with digital subtraction angiography after stent graft deployment.

Episode II

Five weeks later, as he was waiting for discharge and clearance from the hospital, he started bleeding profusely through the tracheostomy tube and re-admitted to ICU. Massive blood transfusions and adequate fluid resuscitation were done on time. An emergent CT carotid angiogram revealed a newly formed right common carotid artery pseudoaneurysm. A second stent graft (Fluency Bard®) 6mm×80mm was deployed to cover the right common carotid artery. Figure 2 The bleeding stopped. No further pseudoaneurysm was seen on digital subtraction angiogram on left/right carotid artery. The intracranial circulation was also intact. After close monitoring in ICU and significant improvement he was transferred to the ward on Day 7.

Episode III

Six weeks later, he re-bled and readmitted to ICU. CT angiogram did not reveal any lesions this time other than a misplaced coil in the neck. Figure 3 Massive bleeding continued overnight, relatives having been counselled and consented to advanced directives of no resuscitation and no intervention based on the guarded prognosis. He passed away the next morning.

Discussion

An aneurysm is a focal dilatation of an artery >50% of the diameter of the natural artery (4). There being no gender predilection, the true incidence of

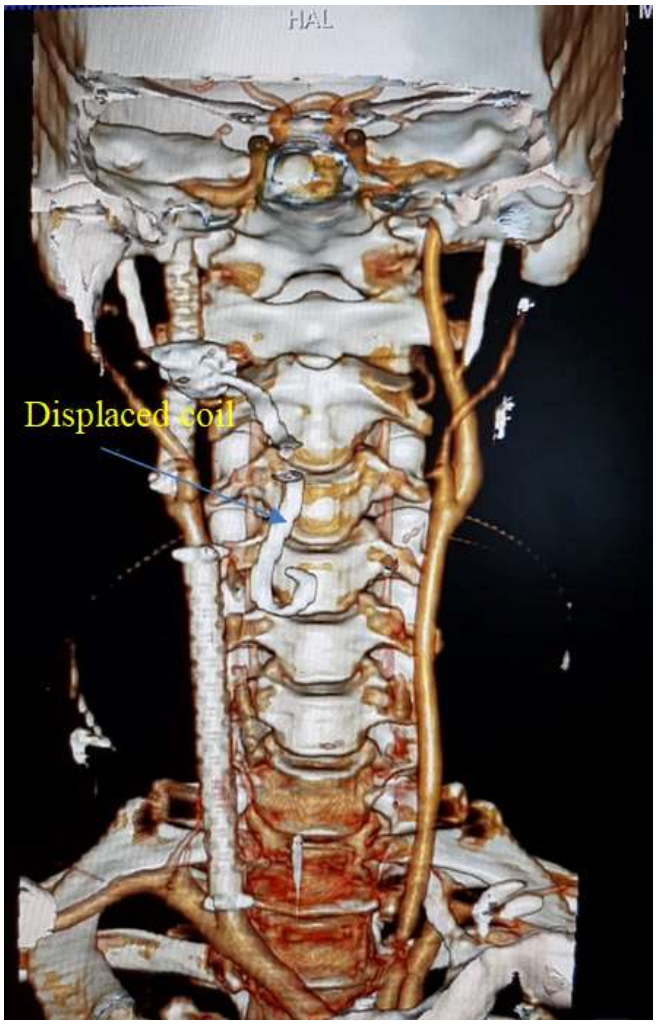

Figure 3. CT angiography images showing stent graft in position, dislodged coil and no new aneurysm as a cause of bleeding.

extra cranial carotid artery aneurysm is probably less than 1% of all carotid disorders (4). Aneurysm of the internal carotid are very rare and pseudoaneurysms of the internal carotid are even rarer.

Pseudo aneurysm occurs when there is an injury of the tunica intima and media, resulting in a hematoma that is contained only by the thin outer adventitial layer of the vessel (5). The absence of a three-layered structure differentiates a PSA from a true aneurysm and makes it prone to rupture more often, unlike a true aneurysm (1)(6) (1,6). Our patient developed multiple PSAs after receiving a course of high dose radiotherapy which has eventually caused friability of the tissues surrounding the neck.

When a patient with massive bleeding is encountered, the algorithmic approach necessitate hemodynamic stabilization of the patient, fluid resuscitation and activation of massive transfusion protocols as the initial step. In our case, timely involvement of an intensivist ensured appropriate measures were taken and the patient had been stabilized. Next step included diagnostic investigations. Clinical examination and duplex

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ultrasound are usually adequate to identify the majority of extracranial pseudo aneurysms.

PSAs can be demonstrated on ultrasound, as a hypoechoic cystic structure adjacent to the true vessel. Duplex doppler ultrasound can demonstrate a "to and fro" color flow and bidirectional Doppler waveform in the pseudo aneurysm neck (7). Although ultrasound was easy to use and noninvasive, it was not used for our patient as the first line of imaging. Instead, CT angiography, which is also safe, fast and accurate tool for diagnosis of pseudo aneurysms, was preferred. CT angiography provides critical information (size, location, neck, collateral circulation and expandability of the artery) which is essential in the choice of the treatment technique.(8)

Hematoma with contrast extravasation seen on angiographic studies is compatible with rupture and active bleeding (7). When a pseudo aneurysm is large enough, it is important to note that the entire sac may not enhance, as the pseudoaneurysm can partially thrombose. In our case, CT revealed both right internal carotid and right common carotid pseudoaneurysms in the first two episodes. MRI with T1-weighted fat-suppressed sequences allows for evaluation of intraluminal thrombus and of pseudo aneurysm sac size. For cases such as our patient with massive oropharyngeal hemorrhage and intractable epistaxis due to extracranial carotid pseudoaneurysms, digital subtraction angiography remains the gold standard for evaluation of pseudoaneurysm and simultaneously offers a therapeutic potential (5)(9), which has been the case for the first two episodes presented here. Embolization and coiling had been attempted by the interventional radiologists, which didn't control the active bleeding and eventually stenting was performed by a vascular surgeon with good clinical and technical outcomes on both times. Hemorrhage was controlled and the pseudoaneurysms were successfully coiled and stented. The coil was later found floating freely in the neck about 3 months after its placement. Endovascular management of extra cranial carotid pseudo aneurysm offers the advantage of avoiding a potentially difficult surgical dissection and eliminating the need for high cervical exposure thus reducing the risk for cranial nerve injuries and other post-procedural complications. Risk of periprocedural stroke and death is very low(9). The low complication and high success rates make endovascular treatment of extra cranial carotid pseudoaneurysm favorable.

In our case, after each successful carotid artery stenting, patient improved with no neurological sequela, ambulating well. Unfortunately, we were unable to prevent the lethal evolution of the disease with recurrent formation of a new pseudoaneurysm.

Conclusions

Multidisciplinary management and endovascular interventional approach involving vascular surgeons, radiologists, ear-nose and throat surgeons, critical care specialists and anesthesiologists is strongly recommended in the management of carotid pseudoaneurysms for effective outcomes. Nevertheless, the caveat is that this condition still carries a significantly high mortality rate.

Conflicts of interests

The authors have no conflicts of interest to declare.

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