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Carotid blowout syndrome: endovascular management of a lesser known oncological emergency: case report

Jitender Singh[1*], Tarika Sharma[2] and Taraprasad Tripathy[3]

Abstract

Background: Carotid blowout syndrome (CBS) refers to a fatal hemorrhagic complication of cervical carotid arteries that occurs due to rupture of the extracranial carotid artery or one of its major branches in patients treated for head and neck malignancy. In this article, we will discuss two different spectrum of CBS and endovascular approach.

Case presentation: Two cases of per oral bleeding presented in the emergency department. After patients were hemodynamically stabilized, CT angiography was done which showed type II CBS and type III CBS, respectively. This was followed by transfemoral supra-aortic digital subtraction angiogram coil embolization with scaffolding and anchoring technique, respectively, for the patients.

Conclusions: Early recognition of the predictors of CBS by a multidisciplinary team is critical. The endovascular treatment approach is relatively safe and effective with low rates of morbidity and mortality compared to surgical approach in CBS.

Keywords: Carotid blowout syndrome (CBS), Endovascular, Coiling, Head and neck cancer

Background

Carotid blowout syndrome (CBS) is a rare potentially life-threatening oncological emergency that occurs in patients treated for head and neck malignancy due to rupture of the extracranial carotid artery or one of its major branches. CBS is categorized into three major types [1]. Threatened (type I) CBS is characterized by carotid artery exposure on physical examination. On vascular imaging, it can be seen as air surrounding the artery with or without abscess, tumor-related fistula or areas of focal arterial wall disruption; however, none of these are characteristic imaging features. Impending blowouts (type II) are sentinel bleeding episodes that can cease temporarily with pressure. On cross-sectional imaging, it is characterized by the presence of a pseudoaneurysm without contrast extravasation. Acute CBS hemorrhage (type III) is life threating, and contrast extravasation is imaging indicator of bleeding point [2].CBS is the result of arterial adventitia ischemia, which can occur following neck dissection with stripping of the carotid sheath, after irradiation for a tumor, by direct tumor invasion of the carotid artery wall, infection and effect of salivary enzymes or multifactorial conditions. The incidence of CBS in head and neck surgery is 3–4.5% [3]. Previous irradiation for head and neck tumors increases the risk of CBS by 7.6 times [4]. Empirical diagnostic CT angiography, catheter embolization and selection of optimal embolic agents can result in optimum life-saving hemostasis.

Case presentations-1

A 52-year-old man with history of smoking and moderate alcohol drinking (no other relevant medical history) was diagnosed with a right retromolar trigone region malignancy, staged as T_3 N_1M_x. He underwent

*Correspondence: introductory2008@gmail.com
[1] Department of Interventional Radiology, Shanti Mukand Hospital, New Delhi, India
Full list of author information is available at the end of the article

chemoradiotherapy treatment (70 Gy of external radiotherapy plus cisplatin). After 6 months of follow-up without any warning signs or symptoms, the patient had an episode of sudden, massive oral hemorrhage at home, subsequently he was hospitalized. Fluid therapy and blood transfusion were given. He was planned for CT angiography as mouth opening was limited to direct endoscopy examination, which depicted large pseudoaneurysm from the right facial artery associated with multiple air foci adjacent to it (Fig. 1a–c). Patient was taken up for DSA coil embolization of facial artery using scaffolding technique (Figs. 1d–f, 2a–c). Results were satisfactory with no residual filling of the sac from contralateral side (Fig. 3a, b). Patient was discharged and is on follow-up in oncology department currently.

Case presentation-2

A 60-year-old male was presented with bleeding per oral in casualty. Examination showed multiple clots in the oral cavity. The patient was in shock. Hemogram revealed hemoglobin of 7 g/dl. Fluid replacement and blood transfusion were given to stabilize the patient. Coagulation parameters were in normal limit. Patient was a diagnosed case of T_3N_2 laryngeal cancer for which concurrent chemoradiation (70 Gy of external radiotherapy and cisplatin) was started a year back which the patient could not follow up. As there was limited mouth opening and patient had severe nausea, nasal intubation was done. After the patient was hemodynamically stabilized, CT angiography was carried out which showed extravasation of contrast adjacent to the right ECA (Fig. 4a–c). The patient was then taken up for transfemoral digital subtraction angiogram (DSA) of neck vessels which showed irregularity of the right ECA wall medially, correlating with site of extravasation on CT angiography (Fig. 4d–f). Following this, the coil embolization with anchoring technique (Fig. 5a–c) was done for which the results were satisfactory (Fig. 6a–c). He was given proper fluid support intravenously and was monitored closely. Patient was

Fig. 1 **a** An axial contrast-enhanced computed tomography (CT) scan demonstrates a mass with obvious enhancement (arrow) in right sublingual space s/o pseudoaneurysm. **b** Coronal section at same level shows osteonecrosis of the adjacent mandible and multiple air foci (arrow) and pseudoaneurysm (open arrow). **c** CT MIP section shows origin of the pseudoaneurysm from right facial artery (arrowhead). Endovascular treatment of facial artery aneurysm. **d** Fluoroscopic image of common carotid artery(CCA) contrast run shows facial artery (arrow) through multipurpose catheter ((MPA catheter, Cook medical llc, USA). **e** Superselective run through microcatheter (2.7 Fr Progreat microcatheter, Terumo, Japan) in facial artery (open arrow) shows pseudoaneurysm sac (arrow). **f** Post coil embolization (hilal embolization microcoil™, Cook medical llc, USA) with scaffolding technique (large diameter coil was first placed forming a 3D scaffold, in that small diameter coil were placed to completely occlude the lumen) fluoroscopic angiogram image shows complete occlusion of feeding artery and preserved distal IMA (internal maxillary artery) and proximal ligular artery

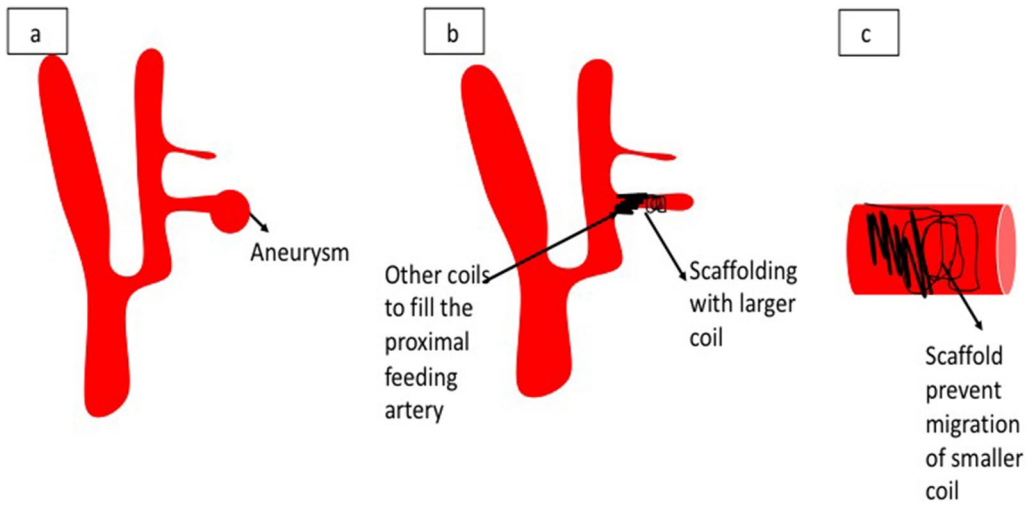

Fig. 2 Graphical representation of scaffolding technique (**a**) representative comparable image with Fig. 1d–f, and shows aneurysm from side branch of external carotid artery. **b** 3D scaffold of large coil in the artery is made and subsequently other coils are placed proximal to it (**c**) enlarged graphical representative image of (**b**) shows close up of scaffolding technique

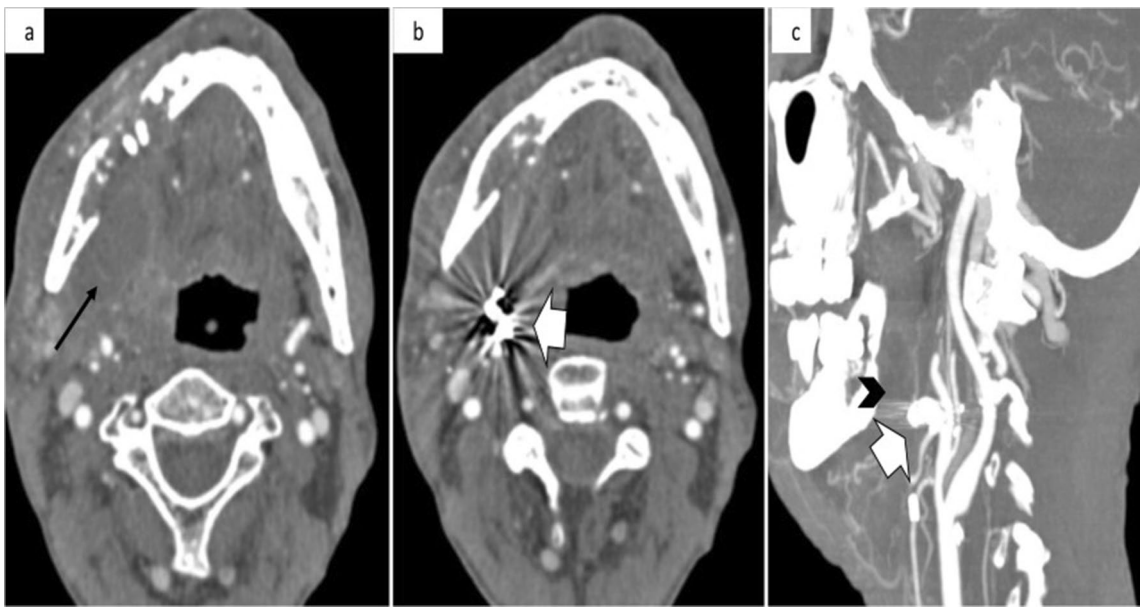

Fig. 3 Post-procedure computed tomography of face; eight days after embolization of case 1. **a** Axial section demonstrates non-opacification of the pseudoaneurysm (indicating successful embolization (arrow). **b** Axial section shows the presence of microcoils (open arrow). **c** Sagittal section MIP image shows a thrombosed aneurysm (arrowhead) and patent rest of arteries except facial artery (open arrow)

extubated and subsequently discharged. Currently, he is on follow-up in the oncology department.

Discussion

We described two patients with head and neck squamous cell carcinoma who were treated with chemoradiotherapy and later presented with per oral bleeding and diagnosed as case of type II and III CBS. Both patients had limited mouth opening due to radiation induced fibrosis which may lead to improper hygiene and oral infection concurrent with low immunity due to chemoradiotherapy. Previous radiotherapy and current oral infection are predisposing factors for carotid blowout syndrome as none of our cases had history of previous surgery or tumor

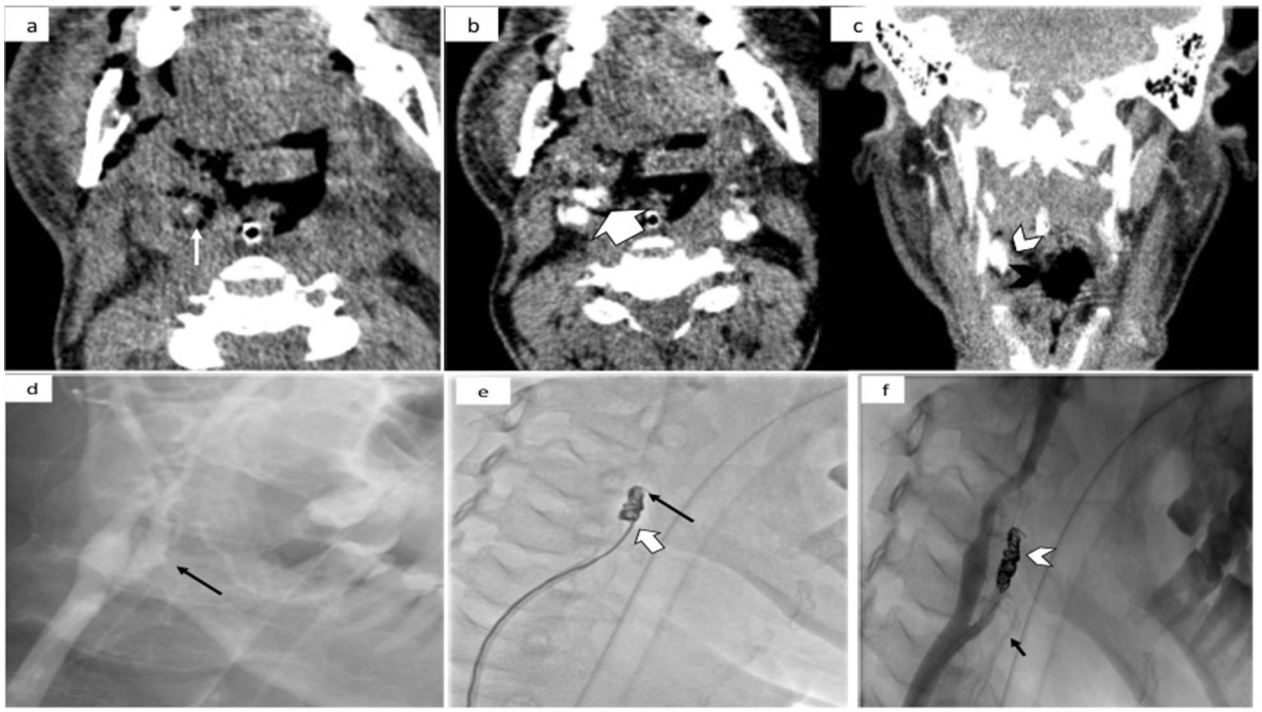

Fig. 4 Computed tomography of patient after stabilization **a** axial non-contrast section demonstrates soft tissue with air foci at the suspected site of external carotid artery breech (arrow). **b** Axial section angiography shows the presence of contrast extravasation (open arrow) and persistence of contrast in delayed image s(not shown here). **c** Coronal CT angiogram shows the presence of contrast extravasation adjacent to right external carotid artery (arrowhead). Endovascular treatment of ECA blowout. Digital subtraction angiography of the same patient. **d** Digital subtraction angiogram through multipurpose catheter (MPA catheter, Cook medical llc, USA) inverted image of common carotid artery (CCA) shows focal outpouching of the right ECA at the suspected site which is correlating with CT angiogram just proximal to ligular artery (arrow). **e** Superselective cannulation of ligular artery using 2.7 Fr Progreat microcatheter (Terumo, Japan) and by anchoring technique (arrow) (long length coil is use to anchor the smaller side branch of the bigger parent artery which help in stabilizing the primary coil in the main artery following which subsequently other coil were placed proximal to that to fill the lumen of the bigger parent artery without migration under the support of the anchor coil) coil embolization (hilal embolization microcoil™,Cook medical llc, USA) of the ECA was done (open arrow). **f** Post coil embolization angiogram image shows complete occlusion ECA distal to superior thyroid artery (arrow) and completer obliteration of ECA and its branches by coil (arrowhead)

invasion on imaging. Both infection and radiation induce vasa vasorum obliteration which may lead to arterial wall thinning and gradual erosion of the arterial wall. In past, surgical ligation has been the only therapy for CBS; however, endovascular approaches have emerged as the standard treatment of choice as these are associated with decreased risk of ischemic neurological events and have lower rates of morbidity and mortality compared to patients who underwent open surgical procedures [5, 6]. Also surgical repair of infected tissue is quite challenging and hence not a feasible option. In both cases, we performed CT angiography first which helped in assessing the vascular anatomy, patency and relationship of the vessels, also in localization of bleeder point, condition of collateral carotids and condition of osseous structures. The previous literature of external carotid artery rupture secondary to osteoradionecrosis has been described in the absence of surgical treatment or tumor recurrence in patients treated with radiotherapy [7]. However, to our knowledge, there have been no previously reported cases of carotid blowout syndrome at facial artery level following chemoradiotherapy. Digital subtraction angiography (DSA) is a gold standard for diagnosing CBS. The main angiographic findings include arterial wall irregularity, luminal stenosis, pseudoaneurysm, arterial pouches and contrast extravasation [6, 8]. However, sometimes angiographic changes can be subtle or even absent, and these unrecognized lesions have high risk of bleeding [8]. Guide wire manipulation is one of the techniques to identify the bleeding point or arterial pouch which are very subtle of imaging [2]. Several endovascular deconstructive and reconstructive techniques including carotids stenting and embolization have been recommended for carotid blowout syndrome. Embolization agents include vascular plugs, coils and permanent balloon occlusion and are commonly used for deconstructive approach. Rebleeding rate in patients who

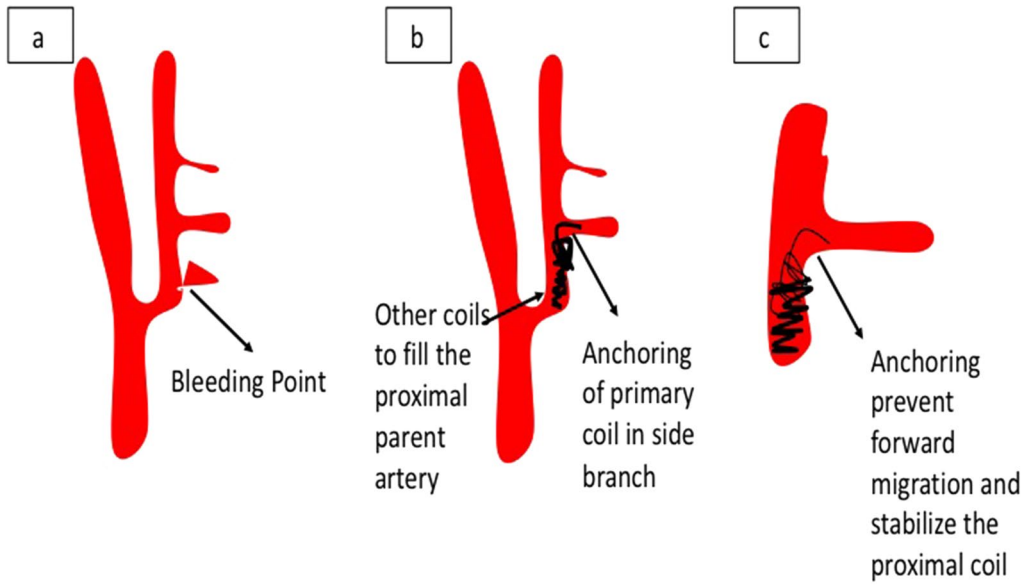

Fig. 5 Graphical representation of anchoring technique (**a**) representative comparable image with Fig. 4d–f shows bleeding point from the main external carotid artery. **b** Anchoring of coil in the side branch of parent artery and subsequently other coils are placed proximal to it (**c**) enlarged graphical representative image of (**b**) shows close up of anchoring technique

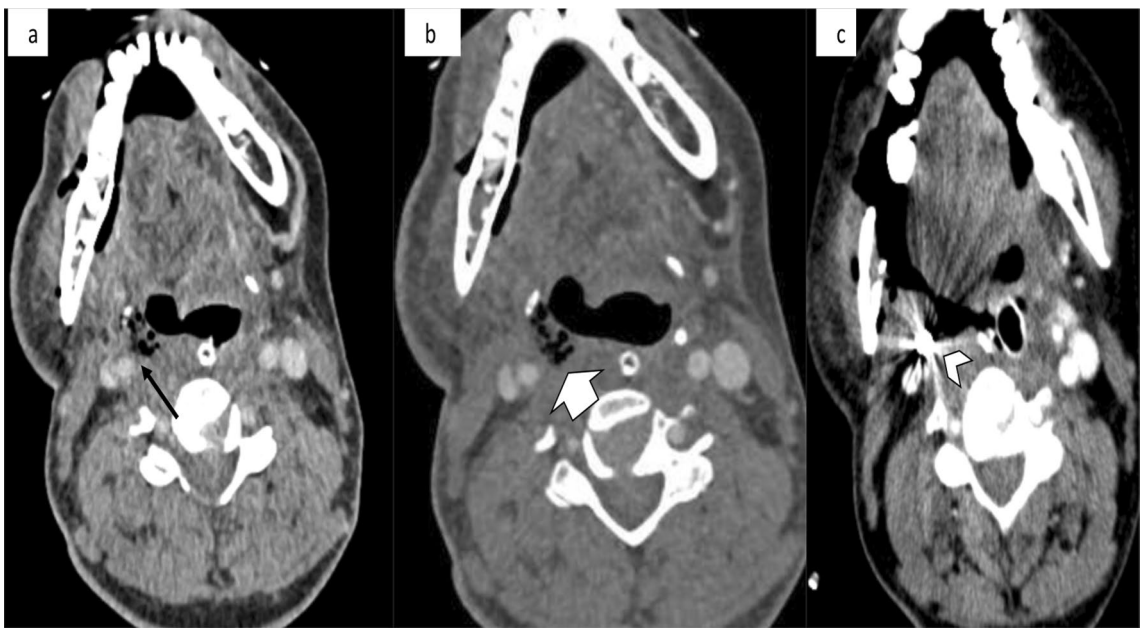

Fig. 6 Post-procedure computed tomography of face 4 days after embolization of case two (**a**) axial section demonstrates no contrast extravasation at the suspected site of external carotid breech indicating successful embolization (arrow). **b** Axial section shows the presence of air foci and no persistence of contrast in delayed section (open arrow). **c** Axial section shows the presence of microcoils in right external carotid artery (arrowhead)

underwent coil embolization is 7% lower than the surgical interventions [5]. Coil embolization using scaffolding and anchoring technique reduces the risk of coil migration contributing to patient safety and lessen the number of coil which ultimately reduce the procedure cost. In scaffold technique which was used in first case, high radial

strength, larger coil with coaxial catheter technique is placed (at least 2–3 mm larger than the lumen) followed by smaller diameter coil into the endoskeleton resulting in complete occlusion of the feeder artery. Clinical severity is the major factor affecting the hemostatic outcome of endovascular management [9]. In anchoring technique, first anchoring coil is introduced with some part into the side branch of the parent artery and some part into lumen of parent artery, and then additional coils are introduced to pack the vascular lumen. In the second case, anchor coil acts as an obstacle to proximal coils against migration.

In the light of our experience, we believe that an active early endovascular approach is associated with higher technical and clinical success.

Conclusions

Early recognition of the predictors of CBS by a multidisciplinary team is critical and the oncologist must be alert to any recent history of per oral bleeding in such patients. In patients with no history of neck dissection surgery, the major risk factors of CBS are infection and radiation therapy. Endovascular techniques are now the standard of treatment in which the proximal bleeder artery embolization with coils is the preferred method. The endovascular treatment of CBS is relatively safe and effective with low rates of morbidity and mortality compared to other treatment techniques.

Abbreviations

CBS: Carotid blowout syndrome; CT: Computed tomography; DSA: Digital subtraction angiogram; ECA: External carotid artery.

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Authors' contributions

All authors have read and approved the manuscript, and data of the cases are shared and ensured by them.

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Availability of data and materials

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Declarations

Ethics approval and informal consent to participate

Not applicable.

Consent for publication

Written consent for publication is obtained from both patients.

Competing interests

None to declare.

Author details

[1]Department of Interventional Radiology, Shanti Mukand Hospital, New Delhi, India. [2]College of Nursing, Institute of Liver and Biliary Sciences, New Delhi, India. [3]Department of Interventional Radiology, Institute of Liver and Biliary Sciences, New Delhi, India.

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