

Contemporary Management of Carotid Blowout Syndrome Utilizing Endovascular Techniques

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Objectives/Hypothesis: To illustrate complex interdisciplinary decision making and the utility of modern endovascular techniques in the management of patients with carotid blowout syndrome (CBS).

Study Designs: Retrospective chart review.

Methods: Patients treated with endovascular strategies and/or surgical modalities were included. Control of hemorrhage, neurological, and survival outcomes were studied.

Results: Between 2004 and 2014, 33 patients had 38 hemorrhagic events related to head and neck cancer that were managed with endovascular means. Of these, 23 were localized to the external carotid artery (ECA) branches and five localized to the ECA main trunk; nine were related to the common carotid artery (CCA) or internal carotid artery (ICA), and one event was related to the innominate artery. Seven events related to the CCA/ICA or innominate artery were managed with endovascular sacrifice, whereas three cases were managed with a flow-preserving approach (covered stent). Only one patient developed permanent hemiparesis. In two of the three cases where the flow-preserving approach was used, the covered stent eventually became exposed via the overlying soft tissue defect, and definitive management using carotid revascularization or resection was employed to prevent further hemorrhage. In cases of soft tissue necrosis, vascularized tissues were used to cover the great vessels as applicable.

Conclusions: The use of modern endovascular approaches for management of acute CBS yields optimal results and should be employed in a coordinated manner by the head and neck surgeon and the neurointerventionalist.

Key Words: Carotid blowout syndrome, carotid reconstruction, endovascular, embolization, and covered stents.

Level of Evidence: 4.

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INTRODUCTION

Carotid blowout syndrome (CBS) represents a devastating complication of head and neck cancer and treatment. The prevalence of CBS for patients treated with advanced head and neck cancer is 3.9%. Risk factors include malnutrition, wound breakdown, pharyngocutaneous fistula, history of radical neck dissection, and total neck radiation dose greater than 70 Gy.^{1,2} The prevalence of CBS may be higher in patients undergoing reirradiation for recurrent disease.³ CBS encompasses three distinct clinical entities: threatened, impending, and acute carotid blowout.² These three entities represent a

spectrum of vessel risk from mere exposure to vessel perforation with acute hemorrhage. Type 1 (threatened) CBS represents vessel exposure with no evidence of bleeding. Type 2 (impending) CBS refers to sentinel bleeding, whereas type 3 (acute) CBS represents overt arterial hemorrhage. CBS carries a high mortality rate, with estimates often exceeding 50%. Relatively less precipitous forms of oropharyngeal hemorrhage can also be a result of tumoral bleeding from branches of the external carotid artery (ECA) and can also be successfully diagnosed and treated with endovascular techniques.

Current management strategies for CBS involve endovascular approaches as a preferred option in centers where such facilities and expertise are promptly available. Endovascular approaches have revolutionized the treatment with a flow-preserving approach (covered stent), or embolization of the bleeding source. Review of nationwide trends shows proportionally higher utility of endovascular sacrifice (88.6%) compared to covered stenting (11.4%) in management of acute CBS. Overall mortality rates (8.0%–10.2%) and acute ischemic stroke rates (2.3%–3.4%) are reported as similar with both strategies.⁴

Endovascular covered stents have been used successfully for management of acute CBS in select cases and provide a window for optimal attempt at definitive vascular reconstruction. However, long-term efficacy of endovascular covered stents for CBS is still under

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TABLE I.
Characteristics of Site of Tumors and Hemorrhage.

Etiology, n = 56	
Head and neck cancer related	33 (58.9%)
Epistaxis	12 (21.4%)
Post-tonsillectomy/biopsy	3 (5.4%)
Diagnostic only	5 (8.9%)
Benign etiology	3 (5.4%)
Age, yr, median (range), n = 33	58 (45–81)
Primary site of tumor, n = 33	
Larynx/hypopharynx	12 (36.4%)
Oropharyngeal	12 (36.4%)
Oral cavity	9 (27.3%)

debate, as there is a requirement for systemic antiplatelet therapy, a high rate of local rebleeding (30%), or conversely, in-stent thrombosis.^{5,6}

In this study, we analyzed the spectrum of oropharyngeal hemorrhage in head and neck cancer patients treated at a single institution and elaborate our management algorithm through the discussion of selected cases.

MATERIALS AND METHODS

Data Collection

After institutional review board (IRB) approval (IRB #08-14-23C) at the University Hospitals Case Medical Center, a dedicated neurointerventional database was used to identify patients treated for extracranial hemorrhage. In addition, patients with a biopsy-proven diagnosis of head and neck squamous cell cancer treated between 2004 and 2014 at the Head and Neck Institute at University Hospitals Case Medical Center and who underwent surgical management of profuse oropharyngeal hemorrhage were reviewed. Patients who were treated for epistaxis, post-tonsillectomy hemorrhage, and bleeding related to benign disease (vascular malformations, endonasal surgery complications) were excluded from the study. Details regarding patient demographics, site of disease, as well as previous surgical and nonsurgical treatment modalities were collected in a retrospective manner.

Embolization and Stenting Procedures

Procedures were performed in a neurointervention suite with monitored anesthesia care using moderate sedation or general anesthesia. Endovascular large vessel occlusion was performed using pushable and/or detachable coils as well as self-expandable occlusion devices (Amplatzer vascular plug; AGA Medical Corp., Plymouth, MN). Flow preservation with covered stent (GORE VIABAHN Endoprosthesis; W. L. Gore & Associates, Newark, DE) was used in selected cases of internal carotid artery (ICA)/common carotid artery (CCA) hemorrhage. In such cases, a postprocedure antiplatelet agent was used to prevent thrombosis. Before parent vessel occlusion of the ICA/CCA, a balloon occlusion test (BOT) was performed in stable patients to assess collateral circulation. In other cases, however, a BOT was not possible because the patient was intubated, sedated, or unstable. In such cases, angiograms of the ICA and vertebral arteries (as needed) was performed bilaterally to evaluate for adequacy of the circle of Willis, so as to estimate potential collateral flow via an anterior communicating artery or posterior communicating arteries. If bleeding was localized to the ECA trunk or its branches, then polyvinyl alcohol (PVA) particles,

pushable fibered platinum coils, detachable coils, or a combination of these materials was used for hemorrhage control.

Descriptive statistics were used to characterize distribution of bleeding, neurological morbidity, and mortality. Survival data were also recorded for the CCA/ICA and innominate bleeding group.

RESULTS

Between 2004 and 2014, 56 patients were identified who were treated using endovascular techniques for bleeding related to extracranial head and neck vasculature. After excluding epistaxis (n = 12, 21.4%), post-tonsillectomy/postbiopsy hemorrhage (n = 3, 5.4%), and other benign etiologies (n = 3, 5.4%), we identified 33 head and neck cancer patients who underwent procedures for control of hemorrhage involving the extracranial head and neck vasculature (Table I). There were five patients who had evidence of clinical bleeding, but diagnostic angiography did not show any discrete evidence of vessel wall irregularity, significant tumor blush, or pseudoaneurysm, and thus no intervention was done. In these five cases, bleeding stopped with conservative management and no further episodes of hemorrhage occurred.

In the 33 remaining patients who ultimately underwent intervention, the median age was 58 years (range, 45–81 years). Distribution of the primary site of cancer is detailed in Table I, with larynx and oropharyngeal sites being the most common. A total of 38 discrete events of hemorrhage were identified in these 33 patients, and all episodes were controlled successfully by the endovascular procedure. Patterns and site of hemorrhage are detailed in Table II.

Twenty-three hemorrhagic events related to distal ECA circulation are also detailed in Table II. Most commonly, the lingual branch of the ECA was involved (47.8% of ECA branch cases), whereas multiple branches were involved in 30.4% of cases and included complex blood supply from the lingual-facial trunk and branches originating from the superior thyroid and inferior thyroid arteries. In all such cases, embolization (coil embolization and/or PVA particles) was used successfully with resultant complete control of hemorrhage.

TABLE II.
Pattern of Bleeding.

Great vessel related, events n = 15	
CCA/ICA	9 (60%)
ECA	5 (33.3%)
Innominate	1 (6.6%)
ECA branches related, events n = 23	
Lingual artery	11 (47.8%)
Facial artery (isolated)	1 (4.35%)
Multiple (lingual-facial trunk, superior thyroid, inferior thyroid)	7 (30.4%)
Superior thyroid artery	1 (4.35%)
Inferior thyroid artery	2 (8.70%)
Internal maxillary artery	1 (4.35%)

CCA = common carotid artery; ECA = external carotid artery; ICA = internal carotid artery.

TABLE III.
Management of Great Vessel Bleeding.

Patient No.	Site of Bleeding/Type of Carotid Blow-Out Syndrome	Etiology	Management	Complications (Neurological)
1	Right CCA (acute)	Recurrent disease	Right CCA embolized	None
2	Left ICA (acute)	Soft tissue necrosis	Left ICA embolized	Permanent hemiparesis
3	Left CCA/ICA (acute)	Pharyngocutaneous fistula	Left CCA/ICA embolized	None
4	Left ICA (acute)	Recurrent disease	Left CCA/ICA embolized	None
5	Right ICA (acute)	Recurrent disease	Right ICA embolized	None
6	Right CCA (acute)	Soft tissue necrosis	Right CCA embolized	None
7	Right carotid (threatened)	Pharyngocutaneous fistula	Right carotid artery stented, embolized after BOT, and eventually resected with closure of wound	Reversible hemiparesis
8	Left CCA (acute)	Soft tissue necrosis	Left CCA pseudoaneurysm stented; left carotid artery resection and reconstruction with superficial femoral vein	Transient aphasia
9	Left CCA (acute)	Soft tissue necrosis	Left CCA embolized	None
10	Innominate (acute)	Recurrent disease	Innominate artery stented	None

BOT = balloon occlusion test; CCA = common carotid artery; ECA = external carotid artery; ICA = internal carotid artery.

Fifteen procedures were related to great vessel hemorrhage including the innominate artery, CCA, ICA, and the ECA trunk. Of these 15 events, five were localized to the ECA trunk, and in all cases successful control was achieved using embolization.

Of the remaining 10 procedures, nine were related to the CCA/ICA, whereas one event was related to the innominate artery. Details of CCA/ICA and innominate bleeding, the treatment employed, and the outcome/complications are depicted in Table III. Nine procedures were performed emergently, whereas one case was elective in the setting of threatened CBS (exposed carotid artery in a case of pharyngocutaneous fistula). Three of 10 cases were managed initially with a constructive (flow preserving) approach utilizing endovascular covered stents with control of immediate hemorrhage. Seven of 10 were managed in a deconstructive approach with sacrifice of the carotid artery. Five of these events were related to unresectable recurrent/persistent disease, whereas the remaining were secondary to soft tissue necrosis/exposure of carotid due to pharyngocutaneous fistula. Most of these patients were transferred from outside hospitals after sentinel events, and the majority had prior radiation treatment elsewhere. Three patients with details on primary radiation had an mean dose of 71.7 Gy. Patients were followed for an average of 16.9 months (range, 0.2–80 months). There was one patient with post-embolization hemiparesis. At the time of study conclusion, four patients were alive, four patients had succumbed to the primary disease, and two patients had an unknown survival status.

Decision Making in Management of CBS

Management of CBS has been revolutionized with the use of modern endovascular techniques. Outcomes including initial control of hemorrhage, rebleeding rates, immediate and late complications, and overall success of

both constructive, flow-preserving, and deconstructive (endovascular sacrifice) approaches have been widely reported in the literature.^{1,7,8}

Our aim in the present study was to highlight complex decision making in these challenging scenarios. Although the goal of initial immediate hemorrhage control is paramount, head and neck surgeons and neurointerventionalists have to identify a myriad of patient- and disease-related factors and employ a strategy that not only ensures rapid control of hemorrhage but also minimizes neurological morbidity and maximizes long-term success. We will discuss approaches for managing acute CBS in the following three scenarios: acute hemodynamic instability, threatened blowout syndrome, and management of acute CBS with contralateral carotid stenosis.

Management of Acute CBS With Hemodynamic Instability

Case 1. A 66-year-old male with prior left oropharyngeal squamous cell cancer treated with definitive chemoradiation presented with a left neck abscess involving the carotid space (Fig. 1A). Direct laryngoscopy and tissue biopsies of the left oropharyngeal necrotic cavity was planned to rule out persistent disease. Intraoperatively, there was acute oropharyngeal arterial bleeding, which was managed with intraoral digital pressure (Fig. 1C). The source was above the carotid bifurcation and based on unfavorable anatomic factors including prior radiation associated rigid neck tissues, active infection, and abscess as well as prior cervical spinal fusion (C4–C6 level), endovascular management was chosen over operative ligation of carotid artery. Figure 1B depicts the digital subtraction angiography (DSA) run postembolization with Amplatzer vascular plugs in the CCA. There was complete and rapid resolution of hemorrhage and no neurological deficits were apparent after extubation.



Fig. 1. (A) Left carotid space abscess abutting the carotid artery. (B) Successful embolization of left common carotid artery with Amplatzer plugs. (C) Focused digital pressure over the site of rupture. Notice the cervical spine hardware that precluded spine extension in this case and hence limited surgical access. (D) Preserved contralateral flow after left vertebral artery injection via a patent posterior communicating artery.



Fig. 2. (A) Exposed right carotid artery in the neck wound. (B) Preserved flow after covered stent placement in the right carotid artery. (C) Endovascular sacrifice of right carotid artery.

Postintervention DSA images via injection of the left vertebral artery showed adequate filling of left middle cerebral artery distribution via a patent posterior communicating artery (Fig. 1D).

Management of Threatened CBS

Case 2. A 52-year-old male presented with recurrent supraglottic squamous cell cancer treated initially with definitive chemoradiation. Salvage laryngectomy and reconstruction with radial forearm free flap was performed. One month after the procedure, he developed a large right pharyngocutaneous fistula with exposed carotid artery (Fig. 2A). He was admitted with concerns for threatened CBS and a covered stent was placed in the right CCA (Fig. 2B). Clopidogrel (antiplatelet therapy) was started. He then underwent successful balloon occlusion testing (BOT). Nutritional and hypothyroid status was optimized and endovascular sacrifice of CCA was then performed (Fig. 2C). Following embolization, he had transient neurological symptoms that completely resolved conservatively. The neck wound was then debrided and eventually closed. He has been doing well with no evidence of recurrence or rebleed 18 months after the embolization procedure.

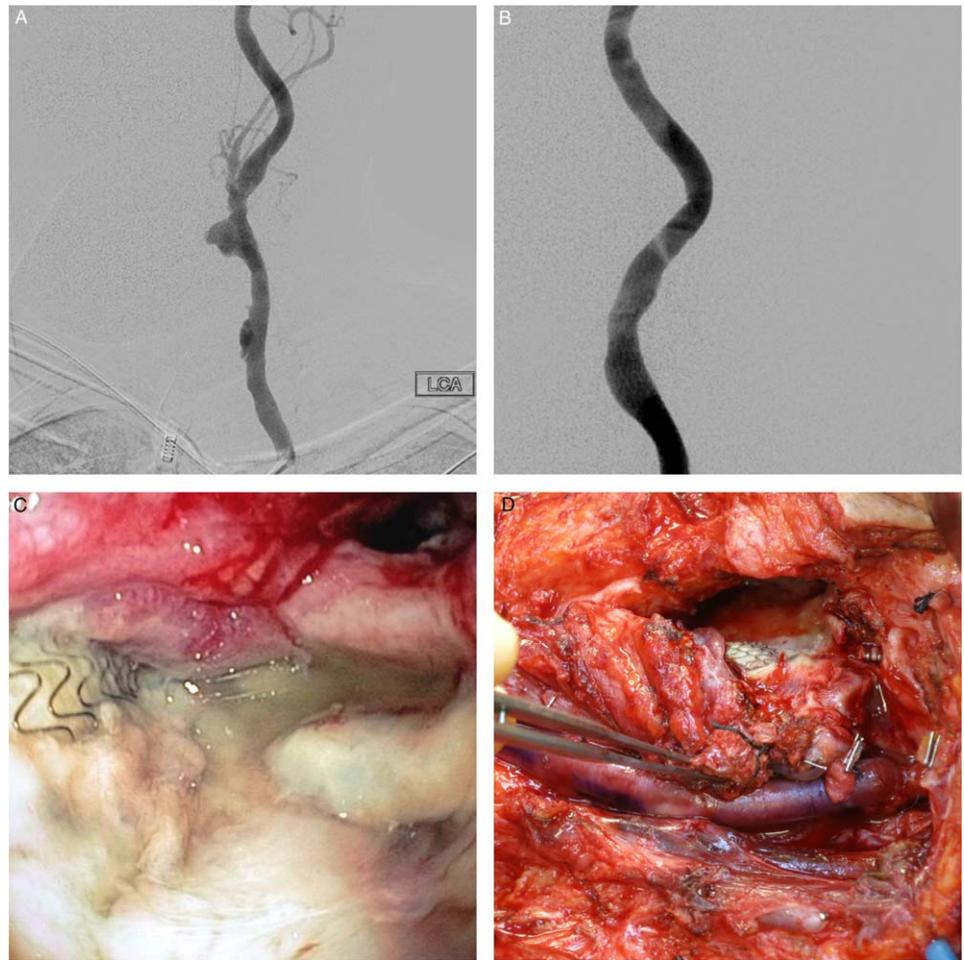


Fig. 3. (A) Left carotid artery (LCA) pseudoaneurysm. (B) Successful placement of covered stent across the neck of pseudoaneurysm. (C) Direct pharyngoscopy showing exposed covered stent in the pharynx. (D) Definitive vascular reconstruction of the LCA with superficial femoral vein graft. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

Management of Acute CBS With Inadequate Contralateral Flow

Case 3. A 63-year-old female with prior history of multiple laryngeal primary cancers underwent salvage laryngectomy for persistent disease. Surgical defect was reconstructed with a pedicled left latissimus dorsi flap. For 19 years she then remained disease free.

In January of 2014, she presented with an episode of sentinel bleed. Computed tomography (CT) of her neck and chest as well as a bedside flexible scope failed to identify any locoregional recurrence or active source of bleeding. Subsequently, during the same admission, she became hypotensive with brisk arterial oropharyngeal hemorrhage. Airway was secured with an endotracheal tube and oropharynx was packed. A massive resuscitation protocol was initiated and she was transferred to interventional radiology.

CT angiography (CTA) demonstrated a pseudoaneurysm of the left CCA just proximal to the carotid bifurcation. There was greater than 50% stenosis of the contralateral ICA. DSA confirmed a left CCA pseudoaneurysm, which was approximately three times the size of a normal CCA but failed to show obvious extravasation (Fig. 3A). Given that there was a contralateral stenosis and that vessel sacrifice would affect the dominant hemisphere, a covered stent (GORE VIABAHN 8 mm × 5 cm)

was positioned across the neck of the pseudoaneurysm and into the ICA (Fig. 3B). The covered stent and ICA were confirmed to be patent. The oropharyngeal packing was removed, and bleeding had resolved. She remained neurologically intact and was discharged 5 days later.

Two weeks later, she presented again with bleeding from a tracheostoma via her tracheoesophageal prosthesis site. Imaging and subsequent direct laryngoscopy showed the recently placed left endovascular CCA covered stent widely exposed in her pharynx (Fig. 3C). Biopsies identified granulation tissue and necrosis but no evidence of recurrent cancer. In anticipation of a left carotid sacrifice a BOT was employed. She failed the BOT and acutely developed significant aphasia and right-sided motor deficits that resolved over the course of 2 days on therapeutic anticoagulation. Once neurologically stable, she underwent reconstruction of her left carotid artery with a superficial femoral vein (SFV) (Fig. 3D) graft and soft tissue coverage with a right supraclavicular fasciocutaneous flap. She tolerated these procedures well and was discharged from the hospital 1 week after her carotid reconstruction. Now, she is 16 months postacute CBS status without any neurological deficit or recurrent disease.

Figure 4 highlights our current management algorithm for management of CBS utilizing modern endovascular approaches as well as surgical control when indicated.

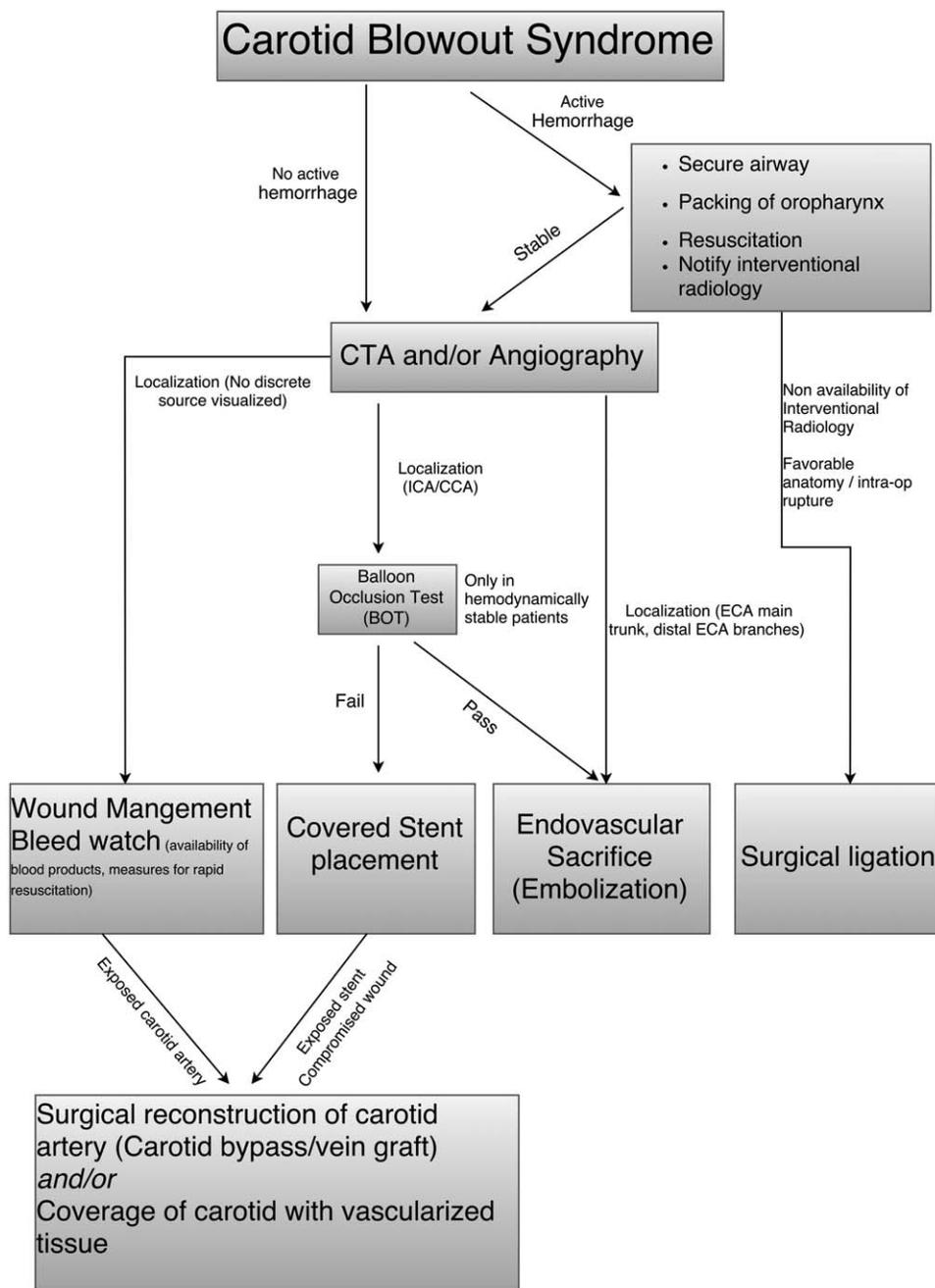


Fig. 4. Proposed algorithm based on type and clinical severity of bleeding. BOT = balloon occlusion test; CCA = common carotid artery; CTA = computed tomography angiography; ECA = external carotid artery; ICA = internal carotid artery.

DISCUSSION

The spectrum of CBS entails challenging scenarios, which require rapid recognition and prompt, often simultaneous, initiation of diagnostic and therapeutic modalities by surgeons and neurointerventionalists. This study affirms the many predisposing risk factors associated with CBS, such as a history of chemoradiation, reirradiation, soft tissue necrosis, tumor recurrence, and poor nutrition.^{2,9} The vast time intervals between treatment and timing of blowout in case 3 emphasize the point that radiation produces long-term tissue changes. This delayed presentation of CBS has been noted in the literature before and may represent only a minor subset of the cases.¹⁰

Surgical ligation for the treatment of hemorrhage related to head and neck cancer historically carried a high mortality rates (40%) and major neurologic morbidity (60%). Over the last 2 decades, the use of endovascular techniques has revolutionized the treatment of CBS with a resultant drop in mortality and neurological morbidity.¹¹ Management of acute CBS involves basic principles of resuscitation with establishment of airway, control of hemorrhage with manual pressure, and/or packing of the oropharynx or neck wound. Rapid resuscitation with blood products and use of other volume expanders/vasopressors is often required as part of initial resuscitation. In patients with threatened CBS, large-bore vascular access and rapid availability of blood products is prudent as well.

CTA can be employed in hemodynamic stable patients to ascertain the likely site of the bleeding and the assessment of patency of the circle of Willis.

If the patient survives this acute hemodynamic change, then utilization of endovascular techniques either in a constructive (covered stent) or a destructive (endovascular sacrifice) manner should be employed based on hemodynamic status, contralateral anatomy, and overall goals, keeping in view the long-term prognosis of individual patients.¹² In the present scenarios, both approaches utilized were successful in achieving immediate control. Reviews have indicated a 95% efficacy rate for endovascular covered stents in controlling acute hemorrhage if the patient can be stabilized long enough to make it to the interventional radiology suite.^{5,6} Although these endovascular techniques may be efficacious for acute hemostasis, their role is often only a temporizing measure. A review of 10 patients with acute CBS, in which endovascular covered stents were placed, showed a 30% rate of recurrent bleeding.⁶ Our experience supports the need for definitive treatment after covered stent placement, particularly if the etiology of the tissue breakdown has not been addressed and is not the result of unresectable cancer. This should be achieved with aggressive wound management and nutritional optimization before rebleeding or stent exposure ensues. In two out of three cases in our series, a covered stent became exposed after a mean duration of 19.5 days. In addition, covered stent deployment in an infected field raises the chances of septic emboli and may serve as a source for potential problems including cerebral abscess.¹³

Definitive management of CBS is dependent upon collateral arterial flow. Case 3 exemplifies this important determinant that guides selection of proper endovascular approach. Initial CTA showed a high-grade contralateral carotid stenosis, indicating that sacrifice of the patient's vulnerable carotid artery would carry increased risk of neurological morbidity, and thus an attempt at reconstruction or bypass must be employed. In this case, the necrotic portion of the CCA and ICA after initial management were resected and reconstructed with a SFV graft. The reconstructed vessels were then covered with healthy, nonradiated tissue from the contralateral supraclavicular region for added protection with no resultant neurological morbidity.

In case of threatened CBS, the imaging and the anatomy should be discussed with the interventional radiologist, and BOT should be done to ascertain collateral circulation. In case 2, we employed an approach of initial covered stenting of the carotid artery followed by nutritional and overall optimization. However, the stent eroded through the arterial wall in the fistula site. Endovascular sacrifice was then employed after the patient passed the BOT successfully. Because of the exposed neck wound, however, the patient had eventual resection of the carotid artery, and the wound was closed with vascularized tissue. This serves to illustrate the critical importance of follow-up, as any exposure of covered stent or coils in the exposed field would require surgical intervention and wound closure.

All stents and particularly covered stents, however, are thrombogenic, and patients require antiplatelet medication prior to the procedure if it is performed electively (i.e., threatened CBS) or during the procedure if it is performed emergently, after ensuring the success of the covered stent in controlling the hemorrhage. Systemic heparinization may also be used in nonemergent procedures and selectively in emergent cases. Furthermore, patients need to be maintained on dual antiplatelet therapy for several months to ensure covered stent patency. Covered stents can also be used for the treatment of pseudoaneurysms of the carotid artery associated with head and neck cancer.

If injury is localized to the ECA trunk or its branches, a BOT is not necessary, because the risk of brain injury during embolization is negligible. PVA particles, pushable fibered platinum coils, detachable coils, or a combination of these materials are used for ECA hemorrhage, which is commonly due to tumoral oozing. There are, however, a number of potentially dangerous ECA-ICA connections that the interventionalist should be aware of when using PVA particles in the external carotid circulation to avoid embolic intracranial complications.¹⁴

CONCLUSION

This study highlights the successful triage and management of CBS using a modern stepwise approach. In patients with impending or acute hemorrhage, utilization of endovascular covered stenting serves to achieve rapid hemostasis while protecting cerebral blood flow. The long-term efficacy of endovascular covered stents largely depends on patient factors such as tissue necrosis or disease recurrence. In most cases, the covered stents should be viewed as a temporizing measure until definitive reconstruction or ablative procedure can be performed. The above case scenarios represent the full spectrum of contemporary, multidisciplinary triage of acute CBS, in which multiple therapeutic interventions were necessary to save the life and limit morbidity.

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