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Case Report

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Emergent Endovascular Parent Artery Occlusion for Type III Carotid Blowout Syndrome After Charged Particle Therapy for Recurrent Maxillary Carcinoma: A Case Report and Literature Review

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ABSTRACT

Carotid blowout syndrome (CBS) refers to involvement of the carotid artery by malignant processes of the head and neck with compromise of vessel integrity and rupture. Carotid hemorrhage (CBS type III) is fatal, particularly when it occurs outside hospital settings. Bleeding can occur through the skin or mucosa and may cause airway compromise. It is associated with high mortality and morbidity. A 55-year-old woman presented to the emergency room with shock state, bleeding from the oral and nasal cavity. The patient had previously experienced recurrent maxillary carcinoma treated with radiation therapy 9 years prior. Digital subtraction angiography revealed active extravasation from the cavernous portion of the left internal carotid artery. Emergent parent artery occlusion was performed by coil embolization, including the extravasation site; complete hemostasis was achieved. Endovascular parent artery occlusion (PAO) is the preferred management method; covered stents are an option for managing patients who are not candidates for PAO. In our case, additional left superficial and middle cerebral artery anastomoses were needed on the day after PAO for preventing hemodynamic cerebral infarction. Since type III CBS can be fatal, early recognition of CBS predictors by multidisciplinary teams is crucial for preventing fatal bleeding.

KEYWORDS: Carotid blowout syndrome, Radiation therapy, Endovascular therapy, Carcinoma, Parent artery occlusion

ABBREVIATIONS: CBS: Carotid blowout syndrome, **PAO:** Parent artery occlusion, **PET-CT:** Positron emission tomography-computed tomography, **RBE:** Relative biological effectiveness, **3D-CTA:** Three-dimensional computed angiography, **DSA:** Digital subtraction angiography, **CCA:** Common carotid artery, **ICA:** Internal carotid artery, **SRS:** Stereotactic radiosurgery, **hSRT:** Hypo-fractionated stereotactic radiotherapy

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INTRODUCTION

Carotid blowout syndrome (CBS) is an uncommon but dreaded complication occurring in patients treated for face, head, and neck cancers. CBS results from arterial wall necrosis. This can occur following resection of face, head, and neck carcinoma, after irradiation of recurrent or second primary tumor, by direct tumor invasion of the carotid artery wall, or by a combination of these factors (14). Advances in interventional neuroradiology have made managing these conditions possible, which was previously difficult (14).

Herein, we present a case of acute type III CBS that occurred in a patient with maxillary carcinoma who actively underwent endovascular emergent parent artery occlusion (PAO).

Written informed consent and consent to publication was obtained from the patient.

CASE REPORT

A 55-year-old woman presented to the emergency room in a serious state with bleeding from the oral and nasal cavity. Examination revealed the presence of large clots in the oral cavity. The patient was in shock. Hemogram revealed a hemoglobin level of 5.1 g/dL. The coagulation profile blood tests revealed normal results. Massive fluid replacement and blood transfusion were performed; however, the patient required continuous blood transfusion to stabilize the blood pressure.

At 37 years of age, the patient had experienced maxillary carcinoma, which was surgically removed and pathologically diagnosed as mucoepidermoid carcinoma. Recurrence was observed on positron emission tomography-computed tomography (PET-CT) 9 years after surgery (Figure 1A, B).

Conformal photon-based planning and charged-particle therapy using charged carbon ions (64 Gy, relative biological effectiveness [RBE]) were used for managing the recurrence. The recurrent lesions were suppressed after radiotherapy (Figure 1C).

The sudden oral and nasal bleeding 9 years after radiation therapy was suspected to be the result of CBS. Emergent three-dimensional computed angiography (3D-CTA) revealed extravasation from the left internal carotid artery (ICA) (Figure 2A, B). An interventional radiologist and neuroradiologist were consulted immediately. Digital subtraction angiography (DSA) revealed active extravasation from the cavernous portion of the left ICA (Figure 2C, D). Emergent PAO was performed by coil embolization, including the extravasation site; complete hemostasis was achieved (Figure 3A-E).

Additional left superficial and middle cerebral artery anastomoses were needed on the day after PAO due to occurrence of a new cerebral infarction in the left ICA watershed region (Figure 4 A-E). The patient was discharged without new neurological symptoms.

DISCUSSION

By definition, CBS is a hemorrhagic event of carotid artery systems (9). It represents involvement of the carotid artery by several lesions of the head and neck (12), causing rupture.

CBS can be categorized into three types that may involve the common carotid artery (CCA) and ICA (8,9,14). Threatened (type I) CBS is characterized by carotid artery exposure detected on examination or imaging in the form of air surrounding the vessel, an adjacent abscess or tumor associated with a fistula, or areas of arterial wall disruption found on vascu-

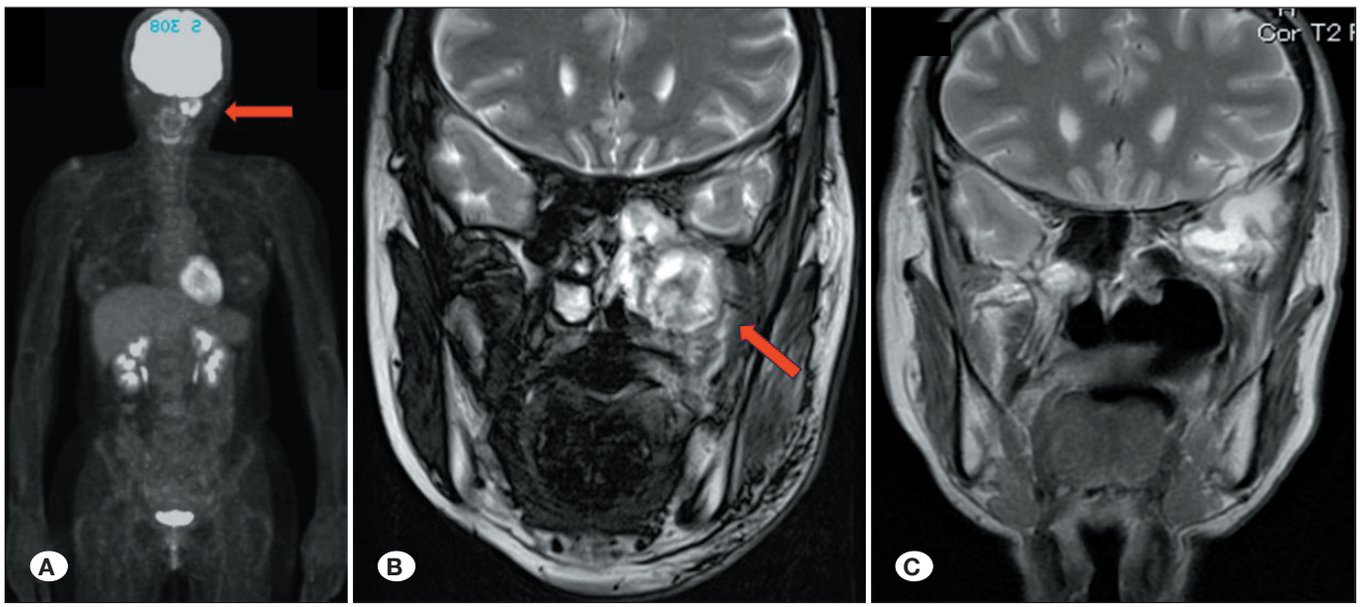

Figure 1: Left maxillary carcinoma recurrence (red arrows) observed in the positron emission tomography-computed tomography **(A),** and magnetic resonance image (MRI) **(B),** 9 years after initial removal. MRI showing suppression of the recurrent lesion after radiation therapy **(C)**.

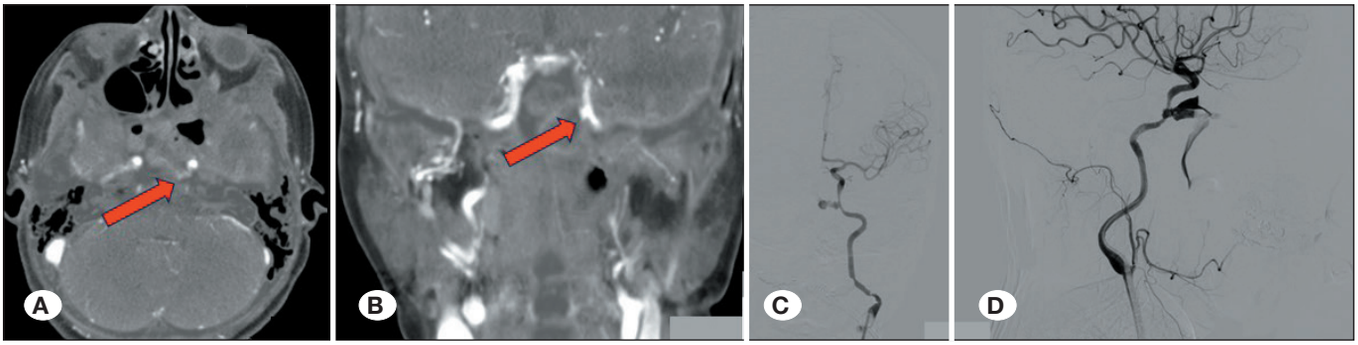

Figure 2: Emergent 3D-computed angiography shows the bleeding from left internal carotid artery (ICA) (red arrows) as suspected (**A:** axial view, **B:** coronal view). Digital subtraction angiogram shows active contrast extravasation from cavernous portion of left ICA (**C:** anterior-posterior view, **D:** lateral view), causing carotid-cavernous fistulae.

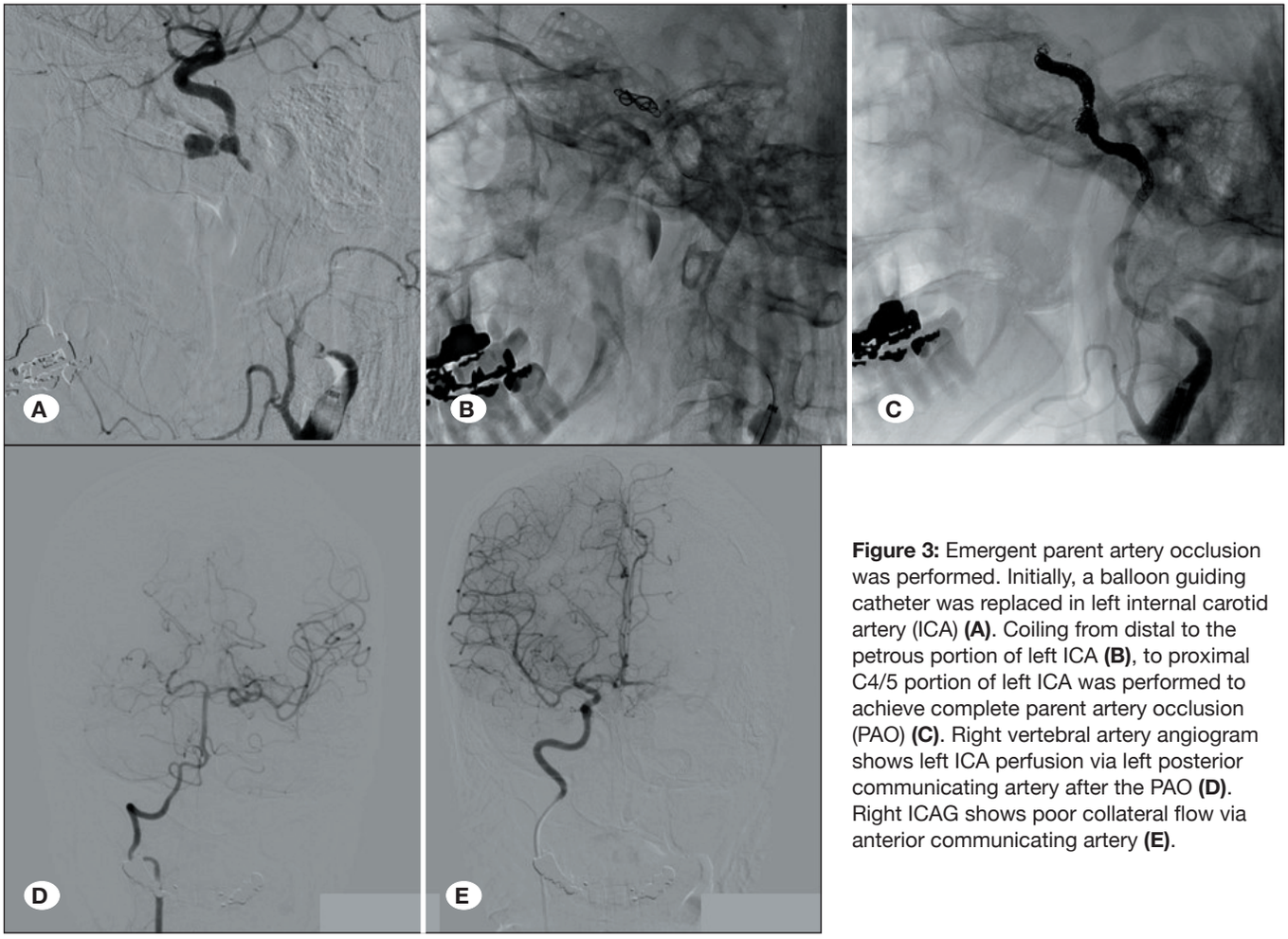

Figure 3: Emergent parent artery occlusion was performed. Initially, a balloon guiding catheter was replaced in left internal carotid artery (ICA) **(A)**. Coiling from distal to the petrous portion of left ICA **(B)**, to proximal C4/5 portion of left ICA was performed to achieve complete parent artery occlusion (PAO) **(C)**. Right vertebral artery angiogram shows left ICA perfusion via left posterior communicating artery after the PAO **(D)**. Right ICAG shows poor collateral flow via anterior communicating artery **(E)**.

lar imaging studies (8,9,14). Impending blowouts (type II) are bleeding episodes that can be temporarily resolved by pressure and wound packing (3). Carotid system hemorrhage (type III) is fatal, particularly when it occurs outside hospital settings (8). Bleeding can occur through the skin or mucosa and may cause airway compromise. It is associated with mortality and morbidity rates of 40% and 60%, respectively (8).

Radiation has been implicated in obliterating the vasa vasorum, causing fibrosis of the adventitia and weakening of the arterial wall, leading to rupture (8,9,14). In the present case, both prior radiotherapy and tumor recurrence were risk factors. Regarding intensity-modulated radiation therapy, the bleeding rates are lower, 0%–2.4%; however, several case series on stereotactic radiosurgery and hypo-fractionated ste-

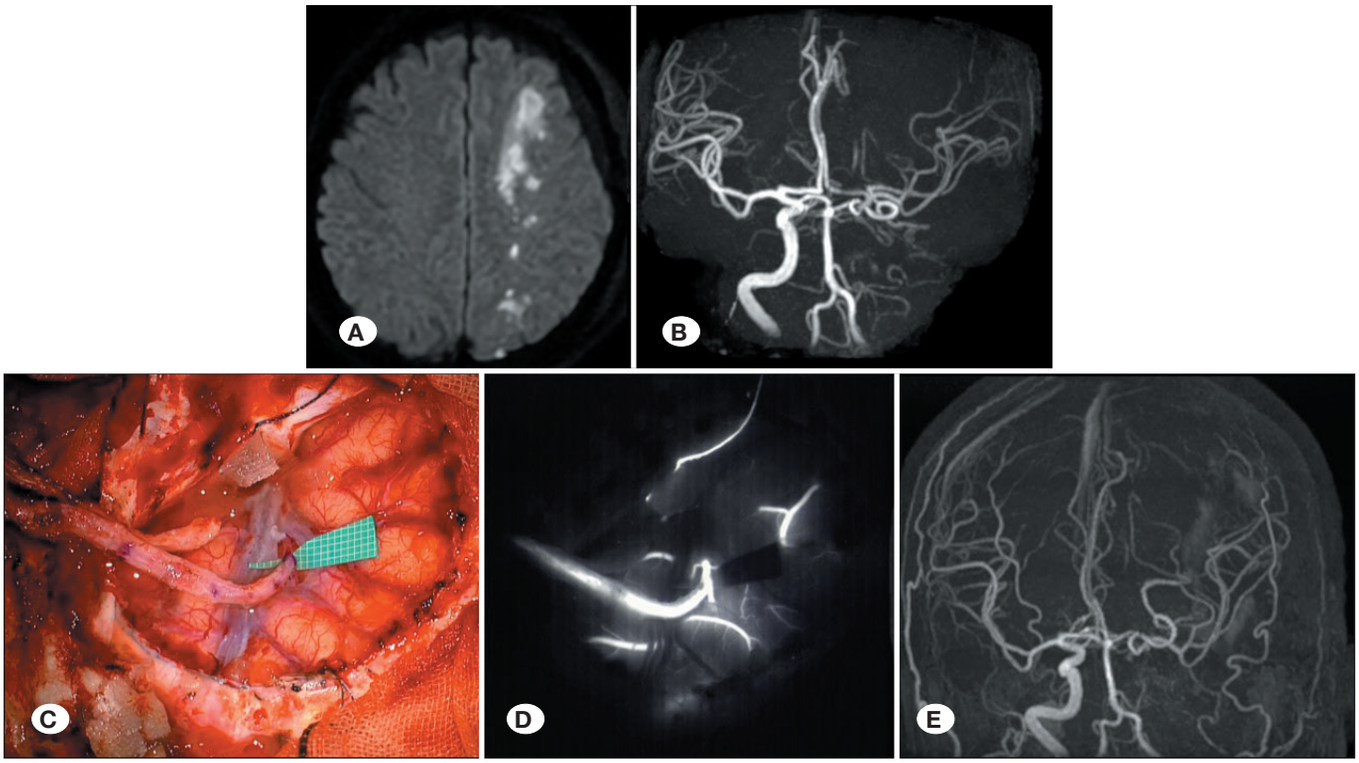

Figure 4: On the next day of the PAO, diffusion weighted image shows high signal intensity in the left internal carotid artery (ICA) watershed region **(A)**. Magnetic resonance angiography (MRA) shows no flow in the left ICA or the extravasation findings **(B)**. Emergent left superficial temporal artery and middle cerebral artery anastomosis was performed for the hemodynamic cerebral ischemia of left ICA occlusion. Intra-operative findings show anastomosis of the superficial temporal artery and middle cerebral artery **(C),** and patent flow on indocyanine green intraoperative angiography **(D)**. Post-operative MRA shows the anastomotic state of the left superficial temporal artery and middle cerebral artery **(E)**.

reotactic radiotherapy have reported higher rates (0–17%) of bleeding (14). Generally, cumulative doses >130 Gy show a higher rate of CBS and other acute and delayed toxic effects than do lower doses (14). In the present case, the patient underwent charged-particle therapy using carbon ions (64 Gy) without chemotherapy and developed CBS type III 9 years after radiation therapy. Charged-particle therapy benefits patients with cancer by localizing doses in the tumor volume while minimizing doses delivered to normal tissue through its unique physical and biological characteristics (6). However, similar to that in our case, late artery rupture can be problematic following this therapy.

The predictors of CBS, such as pseudoaneurysm formation and vessel wall damage should be made aware of after radiation therapy for head and neck malignant tumors (8,9,14). In addition, head and neck surgery was identified as an independent risk factor of CBS in patients post-radiation therapy (4).

Carotid rupture in the setting of re-irradiation in nearly all instances results in death of the patient due to massive hemorrhages in the pharynx or elsewhere. In a systematic review, approximately 80% of cases were fatal, those occurred within one month of CBS onset (5,14). Survival rates at one month and one year after CBS have been reported to be approximately 30% (5,14).

Following establishing a diagnosis of type III CBS, immediate treatment and critical care are essential, including securing the airway and managing hemorrhagic shock (8,9,14). Primary treatment of the ruptured carotid artery mainly comprises endovascular techniques, including embolization and stenting of the parent artery (7), as well as a combination of intracranial and extracranial bypass grafting (2), similar to those in our case.

However, endovascular treatment can induce delayed cerebral ischemic complications, resulting from an incomplete circle of Willis (10). In acute cases, patients have unstable vital signs or impaired consciousness, thereby a balloon occlusion test is usually not possible. Angiography of the contralateral carotid artery and posterior circulation is required to check whether the circle of Willis is complete and whether there is adequate collateral flow, similar to that in the present case. Endovascular embolization of CBS is associated with cerebral ischemic insults in 8%–14% of patients. Although patients at high risk of neurological sequelae are currently treated with covered stents (11,13), cerebral infarction was still reported after such intervention in 15%–30% of patients (14). A higher risk of CBS recurrence with stent placement (44%) has been reported compared with embolization therapy (10%) or surgical ligation (25%) (2). In our case, additional superficial and middle cerebral artery anastomoses were needed on the day

after PAO due to occurrence of a new cerebral infarction in the occluded ICA watershed region. Efficacy of superficial and middle cerebral artery anastomoses for acute ICA occlusion was reported (7). Other complications, both in patients with stents and embolization, include septic thrombosis with multiple brain abscesses, neck abscess formation, and extrusion of the stent or the coils used for embolization (1,5).

CONCLUSION

With the increasing number of treated face, head, and neck cancer cases, oncologists must be cautious about any recent history of oral bleeding or hemorrhage from an exposed neck wound. Once CBS is suspected by an emergency physician, interventional radiologists and/or neuroradiologists must be contacted immediately for expeditious diagnosis and intervention.

PAO is the preferred management method and covered stents are an option for managing patients who are not candidates for parental vascular occlusion. Early recognition of CBS predictors by multidisciplinary teams is crucial.

Declarations

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Availability of data and materials: The datasets generated and/or analyzed during the current study are available from the corresponding author by reasonable request.

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AUTHORSHIP CONTRIBUTION

Study conception and design: DW, TK
Data collection: None
Analysis and interpretation of results: None
Draft manuscript preparation: DW, ST, TK
Critical revision of the article: KM, KS, MN, TT
All authors (DW, ST, TK, KM, KS, MN, TT) reviewed the results and approved the final version of the manuscript.

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