CASE REPORT

Radiotherapy-induced Acute Carotid Blowout Syndrome in the Base of Tongue Cancer

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Abstract

Carotid blowout syndrome is an uncommon and fatal medical emergency in patients with head and neck cancer. Tumor progression, previous surgery, and radiotherapy on the head and the neck regions are some of the predisposing factors that affect its presentation. An early suspicion of a carotid blowout syndrome allows early safety measurements to prevent catastrophic events. A 71-year-old man with advanced base of tongue cancer was presented with a selflimiting oral bleeding 18 months after receiving chemoradiotherapy. Fiberoptic laryngoscopy and contrast-enhanced computed tomography revealed the presence of edema of the hypopharynx and thyroid cartilage fracture and necrosis. The patient developed acute oral hemorrhage. Surgical intervention was insufficient to achieve hemostasis. The carotid angiography revealed a common carotid artery rupture, and a covered stent was placed endovascularly. The covered stent placement was enough to stop the bleeding. However, the patient died from perioperative complications. Experience is necessary for the early detection and prevention of a carotid blowout syndrome.

Keywords

Carotid blowout; Radiotherapy; Covered stent placement

Introduction

A carotid blowout syndrome (CBS) is a rupture of the carotid artery or its branches^[1]. It is a rare and devastating complication of the head and neck cancer (HNC), which is present in 4.3% of the patients, who received radical neck dissection^[1]. Its mortality and morbidity rates are high, approaching 40% and 60%, respectively^[1-3]. It is caused by the cancer, invading the vascular axis or by the cancer treatment method. Prior radiotherapy and surgery are some of the risk factors associated with its occurrence^[4,5]. Carotid blowout syndrome is divided into three types, based on the severity of the bleeding: threatened bleeding, impendent bleeding, and acute bleeding^[2,3]. Acute bleeding is impossible to control without intervention^[2,3]. Surgical intervention is difficult to perform, especially in patients, who have previously received radiotherapy in the head and neck regions. Therefore, endovascular alternatives become the preferable approach in patients with this type of CBS^[6].

This report deals with a case of radiation-induced CBS that was initially presented with a self-limiting,

impending bleeding. The source of the bleeding was not identified through surgical examination, and computed tomography imaging ruled out vascular bleeding. Later, the patient developed acute hemorrhage with an undesirable outcome. Besides the history of receiving radiotherapy, the patient had undergone tracheostomy for the severity of the cancer. Both factors contributed to the development of the CBS in the patient.

Case Report

The 71-year-old male patient had a history of stroke, hypertension, and advanced base of tongue cancer stage IVB (T4aN3M0). He received concurrent chemoradiotherapy with a 60 Gy of external radiotherapy over 30 fractions and 3 cycles of 250 mg/m² of cetuximab. He underwent tracheostomy due to the advanced stage of the cancer, dysphagia, risk of bleeding, and aspiration. The regression of the cancer was confirmed through a surgical examination and a biopsy. An edema in the hypopharynx was found.

Eighteen months later, the patient was presented to the emergency department, with a 6-h history of recurrent self-limiting hemoptysis. He showed stable vital signs and had a preserved hemoglobin level. Fiberoptic laryngoscopy revealed the presence of blood clots in the hypopharynx. However, no active source of bleeding here was identified. Contrastenhanced computed tomography revealed the existence of thyroid cartilage necrosis and fracture (Fig. 1). A severe lumen reduction of the right internal jugular vein with a decrease contrast filling was also observed (Fig. 2). No bleeding from the carotid artery or neoplasm infiltration was noted. The patient was admitted for observation.

Several hours later, the patient had a massive oral hemorrhage with hemodynamic instability. He was transported urgently to the operating room to control the hemorrhage. Under direct laryngoscopy by the otolaryngologist, no active source of hemorrhage was identified, and surgical packing was applied. Shortly after, the patient had another episode of massive

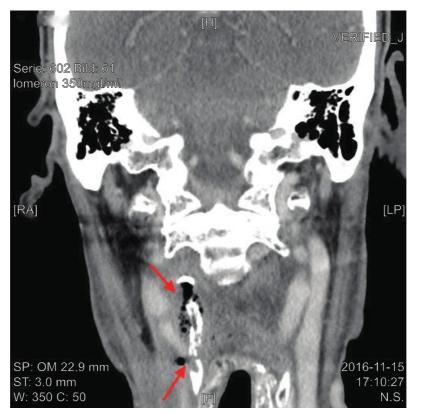


Figure 1. A 71-year-old man with a history of advanced base of tongue cancer, treated with chemoradiotherapy. A contrastenhanced CT of the head and the neck in coronal view shows a fracture line of the right thyroid cartilage (lower arrow), illdefined cortical destruction, and extensive gas bubbles adjacent to the thyroid cartilage (upper arrow). These findings suggest the presence of necrotic tissues.

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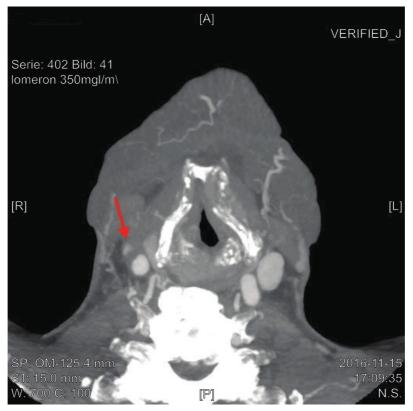


Figure 2. Contrast-enhanced CT with maximal intensity projection shows severe lumen reduction of the right internal jugular vein with decrease in contrast filling (arrow). The possibility of surrounding hematoma cannot be excluded. However, no contrast extravasation was seen.

hemorrhage, and a pulsatile arterial bleeding from the larynx was observed. Aggressive resuscitation with vasoactive drugs, crystalloids, and blood products was needed. The patient suffered from cardiac arrest, and cardiopulmonary resuscitation was immediately initiated. Spontaneous circulation was restored several minutes later. The patient was transferred to the operation room for interventional radiology for an endovascular treatment.

The femoral artery was cannulated. Carotid angiography revealed a 0.5-cm rupture in the right common carotid artery and extravasation (Fig. 3). A Fluency[®] covered stent (Bard Peripheral Vascular, Inc., Tempe, Arizona; code: FEM08040) was placed in the right common carotid artery. Angiography showed good results with no extravasation (Fig. 3). The patient was transferred to the intensive care unit. After several unsuccessful attempts of weaning the patient from the ventilator, he had general myoclonus and signs of severe hypoxic brain injury. Cranial nerve reflexes were absent and scored a 3 on the Glasgow Coma Scale. The patient died shortly after.

Discussion

Carotid blowout syndrome is a rare and a lifethreatening medical condition in patients with HNC, who underwent radical neck dissection^[1]. It gives rise to 40% mortality rate due to massive, uncontrolled hemorrhage that leads to severe hypovolemia and aspiration. Neurological morbidity due to this syndrome approaches 60%, depending on the severity of the cerebral hypoperfusion and the anatomical variation in the circle of Willis^[1-3,7].

Based on the severity of the bleeding, CBS can be divided into three types. The first type is threatened bleeding, where the rupture is imminent without the healthy tissues shielding the carotid artery. It occurs as a result of carotid artery exposure, following a wound breakdown. The second type is impendent bleeding, with self-limiting oral or transcervical bleeding that can be controlled by surgical packing, whenever needed. The third type is acute rupture, which manifests through massive bleeding that cannot be controlled by surgical packing, and it requires surgical intervention^[2,3].

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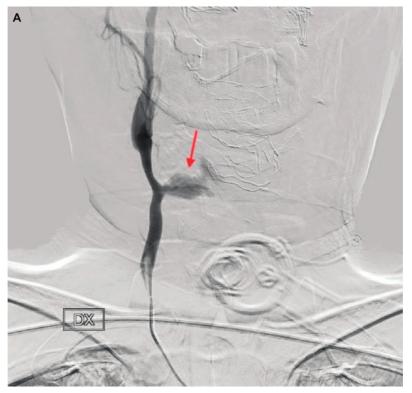


Figure 3. Carotid artery angiogram shows rupture in the right common carotid artery with a contrast extravasation from the medial side (arrow).

Based on literature, CBS occurs as a result of the HNC invading the vascular axis or a postoperative infection and wound breakdown, following a radical neck dissection^[2,5,7,8]. Radiotherapy is one of the most common predisposing factors that underlies CBS, although it is considered an effective treatment option for patients with HNC. Radiation-induced skin ulceration and cartilage necrosis, as in this case, have been reported^[1,8]. Radiotherapy leads to the changes in the vessel wall, including the thinning of the arterial wall, atherosclerosis, and tissue fibrosis, which increases the risk of developing CBS sevenfold^[7,9]. Moreover, one study suggests that surgical or endovascular intervention should be considered in patients with HNC, who had received radiotherapy and were presented with recurrent oral bleeding^[7].

Both surgical and endovascular approaches should be performed to manage CBS. Carotid artery ligation is the conclusive treatment method, but it is associated with higher mortality due to the increased risk of stroke and neurological complications. Radiotherapy leads to vessel changes and anatomical variations, which makes surgical ligation increasingly difficult^[6]. Endovascular treatment, including the placement of covered stent (CSP), coil embolization, and balloon occlusion, has been an attractive method to manage CBS. Covered stent placement has a high success rate of controlling hemorrhage and achieves initial homeostasis. However, it is associated to several complications, including rebleeding, end leak, and stent thrombosis that leads to neurological complications^[6].

Preoperative assessment and perioperative management by a multidisciplinary team that includes otolaryngologist, anesthesiologist, and radiologist are essential^[4]. The potential difficulties in breathing could be due to the tumor that compresses the airway or the tumor that spreads into the airway. Previous radiotherapy treatment is associated with airway fibrosis, the difficulty in ventilation, and the need for intubation^[10]. Computed tomography or magnetic resonance imaging after radiotherapy are required to evaluate the airway patency. Fiberoptic laryngoscopy is a fast and reliable method to guickly evaluate the airway patency in urgent situations. Tracheostomy is performed electively and urgently in different situations with a potential airway compromise.

Adequate airway assessment facilitates planning the approach for patients with HNC, who are opting for surgery, and anticipates the cases with potential difficult airways, as they are at risk of bleeding in the airway and of aspiration that may worsen under general anesthesia due to muscle relaxation and the loss of coughing reflex^[10,11], which was the situation in this patient. Massive blood loss and hemodynamic instability necessitate a collaboration to achieve hemostasis and to ensure adequate organ perfusion^[4]. Optimizing coagulation includes maintaining normothermia, transfusing blood products, and administering coagulation factors^[12]. Invasive blood pressure monitoring and establishing multiple large bore venous access for fluid administration, blood transfusion, and vasoactive drugs administration are essential for the resuscitation of the patient, when the patient is in crisis^[4].

In this case study, the patient had received chemoradiotherapy with complete response that was confirmed with imaging, surgical examination, and biopsy. However, radiation, along with the placement of tracheostomy, had increased the risk of developing CBS. Post-radiation edema and thyroid cartilage necrosis were noted in this patient. In this acute presentation, surgical ligation was contraindicated due to technical difficulties and urgency. Therefore, an endovascular approach was performed. After stent placement, carotid rupture was excluded, and the patient's vital signs were stabilized. Unfortunately, the patient died later as a result of severe brain injury. Despite the unpleasant outcome in this case, the role of the multidisciplinary team was essential to provide the best care possible for the patient.

Brief Summary of the Case

This report relates to the case of a 71-year old male patient with advanced base of tongue cancer, who was presented with adiotherapy-induced CBS. The patient had an impending bleeding, and several hours later, had developed acute bleeding. Angiography revealed the rupture of the patient's right common carotid artery. A covered stent placement stopped the bleeding, but the patient died in the intensive care unit due to perioperative complications. The patient waited for more than six hours before seeking medical help, which has certainly contributed to the outcome of this case.

Conclusions

Experience is necessary to detect the development of CBS in patients at risk, where early intervention should be considered to protect the vascular axis. In an acute condition, a covered stent placement is preferred in order to achieve initial hemostasis. A close follow-up is recommended to detect early complications. More studies are required to evaluate this treatment modality.

A multidisciplinary team is vital in providing such patients with the best possible care. Seeking medical help at the earliest sign of the disease is essential in achieving favorable outcomes.

Conflict of Interest

The author declare that there is no conflict of interest that is related to this study and this article.

Disclosure

The author did not receive any type of commercial support either in the form of compensation or financial support for this case report. The author has no financial interest in any of the products, devices, or drugs mentioned in this article.

Ethical Approval

The study was approved by the Ethics Committee of the KAUH in Jeddah, Kingdom of Saudi Arabia, also known as the Institutional Review Board of Hospitals.

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متلازمة تمزق الشريان السباتي الحاد الناجمة عن العلاج الإشعاعي في سرطان قاعدة اللسان

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المستخلص. متلازمة تمزق الشريان السباتي هي حالة طبية حرجة مميتة غير مألوفة لدى مرضى سرطان الرأس والعنق، و يوجد العديد من العوامل المهيئة للمرض و التي تؤثر على أعراضه السريرية، منها على سبيل المثال تقدم مرحلة السرطان، الجراحة السابقة والعلاج الإشعاعي لمنطقة الرأس والعنق، إذ أن الاشتباه المبكر في متلازمة تمزق الشريان السباتي بناءا على استيفاء هذه العوامل، يساعد على اتخاذ إجراءات السلامة في وقت مبكر لمنع حدوث مثل هذه الكارثة - والحالة لرجل يبلغ من العمر ٢١ عاما مصابا بمرحلة متقدمة من سرطان قاعدة اللسان، مصحوبًا بنزيف فموي بعد مضي ١٨ شهرًا من تلقيه العلاج الكيميائي والإشعاعي حيث أظهر تنظير الحنجرة والتصوير المقطعي باستخدام الصبغة وجود أورام في البلعوم، كما كشف عن وجود نخر وكسر في الغضروف الدرقي، و أصيب المريض بنزيف فموي حاد وكان التدخل الجراحي يلياف النزيف.

كشف تصوير الأوعية الدموية تمزق الشريان السباتي المشترك، وتم وضع دعامة مغطاة داخل الشريان من داخل الأوعية الدموية، و كان استخدام الدعامة المغطاة كافيا لإيقاف النزيف، غير أن المريض انتقل إلى رحمة الله متأثرا بمضاعفات متعلقة بالجراحة، فوضع الدعامات المغطاة هو وسيلة فعالة لعلاج هذه الحالة، ولكن ينبغي النظر في طرق العلاج الأخرى كما أن الخبرة ضرورية للكشف المبكر، والوقاية من متلازمة تمزق الشريان السباتي.