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CASE REPORT

Endovascular intervention in hemorrhage from oropharyngeal tumors: Case presentation of embolization of the lingual artery with coils

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Oropharyngeal tumors represent a multidisciplinary challenge in terms of localization, potential local invasion, and associated symptoms. When patients present with active bleeding from the tumor, endovascular intervention may be essential to control the bleeding and stabilize the patient. Lingual neoplasm is a complex entity associated with significant morbidity, especially when it progresses with ulceration. In cases where the oropharyngeal tumor presents with active hemorrhage, endovascular embolization is an important therapeutic option for immediate hemorrhage control, thus contributing to hemodynamic stabilization and allowing the initiation of oncological treatment strategies.

This case report presents a 60-year-old chronic smoker with a known lingual neoplasm who presented to the emergency department with active oropharyngeal hemorrhage. On examination, the patient showed signs of significant bleeding from the base of the tongue and had an ECOG-PS of 3 (Eastern Cooperative Oncology Group Performance Status). Immediate intervention was necessary to control the bleeding and prevent further complications. After initial stabilization, the patient underwent angiographic evaluation, which identified active bleeding from the right lingual artery. Following the failure of conventional hemostatic methods, selective embolization of the lingual artery with permanent coil embolization material was decided. Post-procedural monitoring confirmed the absence of bleeding.

This case highlights the importance of prompt and precise multidisciplinary management of complex cases, which has a significant impact on patient outcomes. Initial management focused on hemorrhage control, which could impede the use of conventional oncological therapy known for its potential to negatively affect the healing process and tissue integrity. As an alternative to invasive surgical treatments and as a preliminary step in transitioning to oncological therapy, this paper recommends embolization as a valuable therapeutic option in similar circumstances. By effectively controlling hemorrhage, this procedure allows oncologists to proceed with cancer treatment strategies, minimizing delays and reducing the risk of further complications. Given the high incidence of smoking, increasing awareness and the use of these advanced interventional techniques are crucial for improving outcomes and reducing associated complications.

Keywords: lingual hemorrhage, hemorrhagic complication, interventional radiology, arterial embolization, superselective catheterization, coil embolization, hemostasis

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Introduction

Base of tongue cancer represents a category of oropharyngeal cancers with a poor prognosis in advanced stages. The most common etiological factors are human papillomavirus infection, excessive alcohol consumption, and smoking. The main characteristics are extensive loco-regional invasion and complications such as hemorrhage and upper airway obstruction [1]. Lingual artery embolization is a widely used technique in various medical scenarios, including the treatment of bleeding pseudoaneurysms of the lingual artery, reduction of perioperative bleeding during transoral robotic surgery, management of acute oral hemorrhage and hypovolemic shock caused by small pseudoaneurysms of the lingual artery, and control of bleeding in patients with unresectable, inoperable, recurrent, and/or metastatic head and neck cancer. Studies have demonstrated the success of preoperative transarterial embolization for solitary fibrous tumors of the tongue and the use of embolic agents for bleeding in non-neurological procedures and carotid blowout syndrome associated with head and neck cancer. The importance of this technique in achieving favorable outcomes and reducing the risk of complications in various clinical scenarios is well documented in the literature [2,3]. The first embolization agent used in 1974 was made from autologous blood clot. Embolization agents, whether temporary or permanent, are substances or materials used to obstruct blood flow. Embolization agents are classified into two main categories based on the duration and purpose of their effect. Temporary embolization agents include autologous blood clot, gelatin sponge or powder, starch microsphere, microfibrillar collagen hemostat, and phase-change material-based nanoparticles such as thrombin. These are used to reversibly occlude blood vessels, useful in clinical cases requiring subsequent restoration of flow. Permanent embolization agents include microparticles such as polyvinyl alcohol (PVA), coils (implantable, injectable, detachable), liquid agents (cyanoacrylate adhesives like Glubran, Onyx, absolute alcohol, sodium tetradecyl sulfate, alginate), devices like Amplatzer plugs and balloons, and subcutaneous tissue. These are used to create a durable occlusion [4].

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Case presentation

We present the case of a 60-year-old patient with a known history of lingual neoplasm who presented to the emergency department for imaging evaluation and specialized treatment for oropharyngeal hemorrhage, dysphagia, significant weight loss, asthenia, and extreme fatigue. The patient's history includes chronic smoking and alcohol consumption, and a previous hospitalization in the pneumophysiology department due to chronic smoking-related cough, anorexia, and weight loss. Investigations revealed the presence of apical pulmonary nodules and adenopathies, raising suspicions of systemic involvement, possibly neoplastic. Although pulmonary pathology was not confirmed as malignant, chronic inflammation and a potentially compromised immune status may have contributed to immunosuppression and susceptibility to developing lingual neoplasm.

Two years later, the patient underwent extracapsular cataract extraction and artificial lens implantation. The patient is known to have chronic obstructive pulmonary disease (COPD) and chronic liver disease, for which therapeutic recommendations were not followed. Cachexia due to inadequate nutrition and existing comorbidities limit therapeutic options, complicating the prognosis and management of the disease.

Six months prior to the current admission, following an oromaxillofacial consultation, additional clinical and imaging investigations were performed for a painful neoplastic formation at the base of the tongue. The histopathological examination of a specimen from the marginal lingual tumor revealed, macroscopically, a 5/5/3 mm brown-gray elastic fragment, entirely included. Histopathological diagnosis: fragment of fibroconnective tissue with the presence of mucous glands and a rich inflammatory infiltration. At one margin, there is a hyperplastic stratified squamous epithelium without dysplastic changes. The examined fragment does not indicate the presence of specific malignancy characteristics, such as tissue invasion or severe dysplastic changes. It is important to note that the absence of dysplasia does not completely exclude the possibility of a very early stage of malignancy or a benign process.

The objective examination revealed the patient's general condition is mediocre, with an ECOG-PS of 3, indicating significantly reduced functional capacity. The skin and mucous membranes are pale and dry, and the face has a suffering appearance. The connective-adipose tissue is diminished, and the superficial lymphatic system is not palpable. The muscular system shows normal tone and kinetics, and the osteoarticular system appears intact. The thorax is normally shaped, with physiological vesicular murmur, and blood oxygen saturation (SaO2) is 97%. Cardiac examination reveals rhythmic sounds, with a ventricular rate of 80 beats per minute and blood pressure of 120/70 mmHg. The abdomen is non-tender both spontaneously and on palpation, and intestinal transit is present. Swallowing is impossible for both liquids and solids. The patient presents anorexia and expectorates blood-tinged sputum. Urination is physiological. The patient is temporo-spatially oriented, osteotendinous reflexes are present, and the emotional state is characterized by anxiety.

Laboratory results show the following deviations from normal ranges (the normal ranges provided are those established by the laboratory that analyzed the results).

Elevated values:

- Potassium: 5.29 mEq/L (normal range: 3.5-5.1 mEq/L)
- Platelet count: 451 x 10^3/µL (normal range: 150-400 x 10^3/µL)
- Plateletcrit: 0.39% (normal range: 0.17-0.35%)
- Monocytes: 16.3% (normal range: 2-10%)
- Eosinophils: 4.2% (normal range: 1-4%)

Decreased values:

- Sodium: 133 mEq/L (normal range: 136-145 mEq/L)
- Chloride: 97.8 mEq/L (normal range: 98-107 mEq/L)
- Red blood cell count: 3.25 x 10^6/µL (normal range: 4.5-5.8 x 10^6/µL)
- Hemoglobin: 9.8 g/dL (normal range: 13-17 g/dL)
- Hematocrit: 30.6% (normal range: 40-50%)
- Mean platelet volume: 8.7 fL (normal range: 9-13 fL)

The patient presents signs of anemia indicated by low values of red blood cells, hemoglobin, and hematocrit, thrombocytosis, and an imbalance in the components of the serum ionogram profile.

The MDCT (multi-detector computed tomography) cervico-thoracic native and post-contrast intravenous examination with contiguous sections, under conditions of extremely difficult venous access with slow contrast injection, reveals, in the absence of fatty demarcation planes, an iodophilic tissue mass, ulcerated at the median and right paramedian lingual level, approximately 4/4.8/5 cm in size. This mass extends towards the right palatine tonsil and the right paramedian floor of the mouth, causing asymmetric narrowing of the oropharyngeal lumen and a tendency to fix the uvula (Figure 1). Distally, it occupies the right vallecula and right piriform sinus, lyses the right posterior horn of the hyoid bone, with subcutaneous bulging at this level. The described mass encompasses the initial portion of the right external carotid artery (ECA), with absent flow at this level. No pathological adenopathies are detectable on CT at the laterocervical level. There is a fibrotic sequela area in the right apical lung with a small thin-walled cavitary lesion approximately 1.7 cm in diameter. The pulmonary interstitium is more pronounced basally on both sides. There is a lateral left lower lobe pulmonary micronodule approximately 0.6 cm in size. The aorta is ectatic with a diameter of 4.6 cm. No mediastinal adenopathies or pleural collections are present. No pathological changes are detectable on CT at the upper abdominal level.

The imaging examination concludes with an iodophilic lingual tissue mass, median and right paramedian, ulcerated, with loco-regional invasion, a sequela area with a small

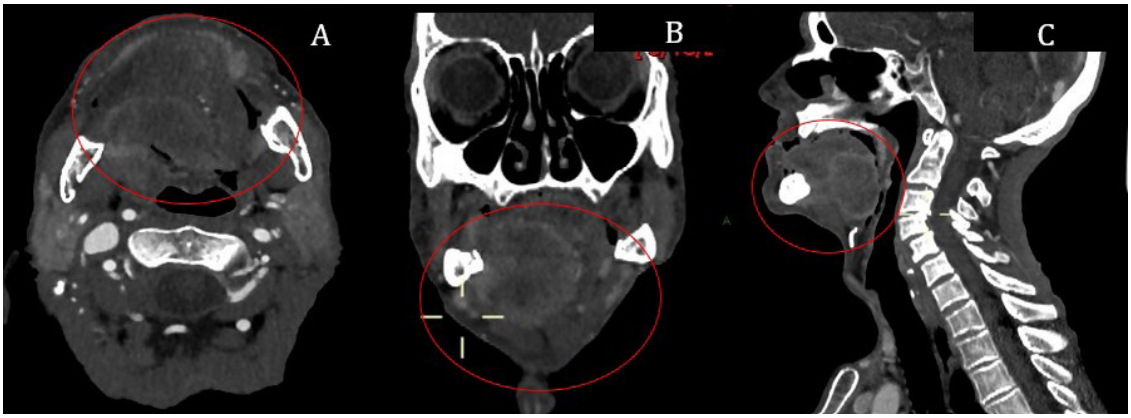

Fig. 1. Multidetector computed tomography sections in different cross-sectional planes: transverse (A), coronal (B), and sagittal (C), with the indication of iodophilic mass

cavitary lesion in the right upper lobe apex, and a lateral left lower lobe pulmonary micronodule.

The MDCT examination reveals an extensive and invasive tumor mass at the level of the tongue and adjacent structures, involving vital structures such as the right external carotid artery and the hyoid bone, strongly suggestive of an advanced malignant process. In the pulmonary region, chronic interstitial changes and nodules were identified, necessitating further investigations to exclude the possibility of pulmonary metastases. Additionally, aortic ectasia may indicate an aneurysmal dilation, requiring further monitoring due to the risk of rupture. Due to the active and persistent bleeding from the right hemilingual region, the patient was directed to the Interventional Radiology Service.

Embolization – Descriptive pathway of the minimally invasive approach

The actual embolization technique consists of two distinct stages: a diagnostic phase and a therapeutic phase. In the first stage, the diagnostic component, the exact source of the hemorrhage is identified through selective angiography, allowing precise visualization of the involved lingual artery. Subsequently, during the therapeutic phase, the actual embolization is performed by placing a coil to obstruct blood flow and control the hemorrhage. Detailed and in-depth knowledge of the lingual vascularization and the territories it irrigates is essential for the correct approach to associated pathologies.

The lingual artery moves anteriorly and anterosuperiorly, passing deep to the posterior belly of the digastric muscle and the hyoglossus muscle. It inserts on the greater horn of the hyoid bone and the lesser horn through the two portions of the ceratoglossus and chondroglossus muscles. The lingual artery continues medially to the posterior belly of the digastric muscle and deep to the hyoglossus muscle, emerging from under it and heading towards the tip of the tongue. The artery is located deep to the inferior mucosa of the tongue and terminates through its terminal branch, the deep lingual artery. The lingual artery, a major vessel responsible for irrigating the tongue, presents several branches with distinct functions and distributions as it progresses towards the tip of the tongue from the external carotid artery. The main branches are: the suprahyoid artery, located superior to the horn and body of the hyoid bone with the possibility of anastomosing with the one on the opposite side; the dorsal lingual arteries, consisting of three thin branches that vascularize the posterior third of the tongue, the palatoglossal arch, the lingual tonsils, the soft palate, and the epiglottis; the sublingual artery, which vascularizes the sublingual gland, genioglossus, geniohyoid, mylohyoid, buccal mucosa, gingival mucosa, and mandible; the deep lingual artery, the terminal branch also known as the ranine artery, which vascularizes the body and tip of the tongue. The diameter of the lingual artery varies on average between 2-5 millimeters and increases as it approaches the external carotid artery [5-7].

During the diagnostic phase, the right femoral artery was punctured under local anesthesia with lidocaine, which was buffered with sodium bicarbonate. Buffering the local anesthetic with sodium bicarbonate reduces the acidity of the solution, leading to decreased pain and discomfort upon injection. The anesthetic effect is accelerated by increasing the pH, and its efficiency is enhanced by the persistence of lidocaine molecules in their active form due to the less acidic environment [8]. Local anesthesia for femoral artery puncture is administered in the inguinal region, below the inguinal ligament, at the level of Scarpa's triangle, approximately halfway between the anterior superior iliac spine and the pubic symphysis, where the common femoral artery is superficial and accessible. After puncture, a 5F (French) femoral introducer is placed. Selective catheterization is performed by choosing a catheter with a curvature suitable for the emergence of the artery to be catheterized. Under angiographic guidance, the guidewire and catheter are advanced through the femoral artery to the external iliac artery, common iliac artery, abdominal aorta, thoracic aorta, and up to the level of the aortic arch. From there, they are directed towards the brachiocephalic trunk, which branches into the right common carotid artery and right subclavian artery. The guidewire continued along the right common carotid artery, and to reach the

lingual artery, it traversed the external branch of the carotid artery. In this case, a 5F angled/vertebral diagnostic angiographic catheter was used for selective catheterization of the right vertebral artery, right and left common carotid arteries, and external carotid artery. After performing angiographic series in frontal and lateral projections (Figure 2), the vertebral catheter was placed at the origin of the right external carotid artery.

During the therapeutic phase of the intervention, superselective catheterization of the right lingual artery was performed using a microcatheter (Figure 3 A). The variable diameter of the lingual artery and its branches, which vascularize different segments of the tongue, justify the use of a microcatheter with a small diameter, usually between 0.4 and 1 mm, for superselective catheterization. This allows precise access to small arterial branches, minimizing disruption of blood flow.

The coaxial system involved the use of a main catheter and a guidewire, which were positioned to guide the microcatheter. The microcatheter was inserted through the lumen of the main catheter and selectively advanced to the collateral branch of interest. Superselective embolization was achieved by the precise delivery of the embolic material, in this case, simple coiling, thus occluding the hemorrhagic source at the base of the right hemilingual (Figure 3 B, C). After delivering the permanent embolic material, the instruments were withdrawn, and the femoral puncture site was closed by manual hemostasis through compression.

The embolization of the right lingual artery using simple coiling was performed without procedural complications. To evaluate the impact of the intervention on the entire lingual vascular system, angiography of the left common carotid artery was also performed (Figure 3 D). This allowed visualization of the left lingual artery and assessment of blood flow, which, through collateral vessels, could have compensated for the obstruction of the right lingual artery. Additionally, it was possible to evaluate any changes in blood flow that could have influenced the patient's clinical outcome and to rule out the possibility of another bleeding source. The angiographic series performed at the level of the two external carotid arteries excluded the presence of active bleeding. In conclusion, angiography of the contralateral homonymous arteries completed the post-embolization evaluation.

Discussion

Embolization with simple coiling of the lingual artery to control hemorrhage in a patient with lingual neoplasm involving the initial portion of the external carotid artery can be a life-saving procedure, with clinical outcomes that can ameliorate post-hemorrhagic complications, especially in the context of anemia syndrome associated with neoplastic disease. However, it involves significant risks.

Post-procedural monitoring is essential to detect post-embolization syndrome, requiring increased attention to symptoms that may appear within the first 24 hours (fever, nausea, vomiting). It is important to administer appropriate supportive treatment, including antipyretics, analgesics, and antiemetics, as well as careful evaluation of possible infections or local ischemia.

Among the major complications associated with lingual neoplasm involving the external carotid artery that require attention are:

- Lingual Ischemia: Resulting from the interruption of blood flow post-embolization, which can lead to tissue necrosis. Observable signs and symptoms include lingual edema accompanied by severe pain, tissue necrosis that may require debridement or glossectomy, and speech and swallowing disorders due to loss of muscle function [9];
- Hypoglossal (XII) and Glossopharyngeal (IX) Nerve Damage: Due to mechanical manipulation during the procedure or secondary ischemia, with conse-

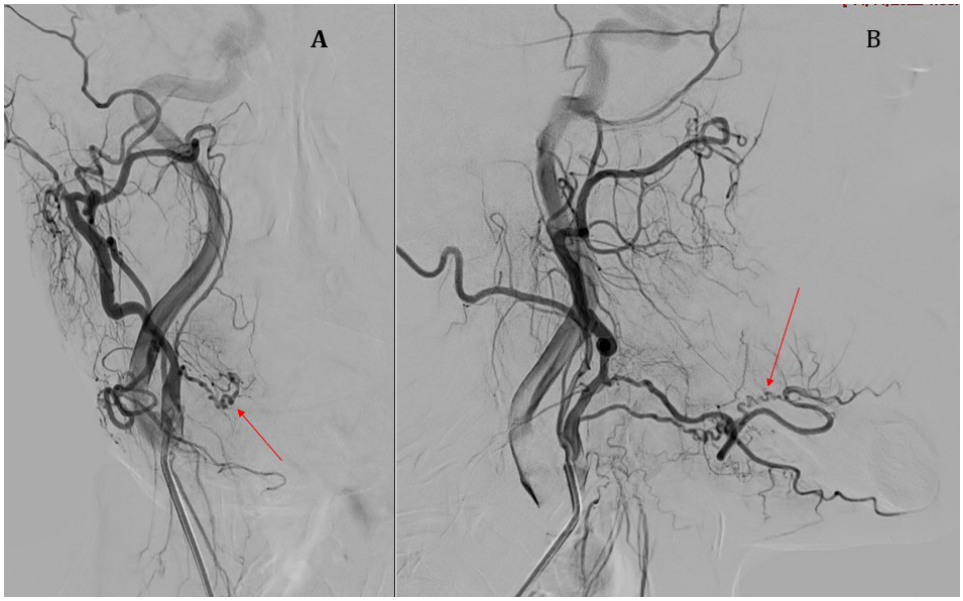

Fig. 2. Global arteriography in anteroposterior (A) and lateral (B) projections, highlighting the lingual artery

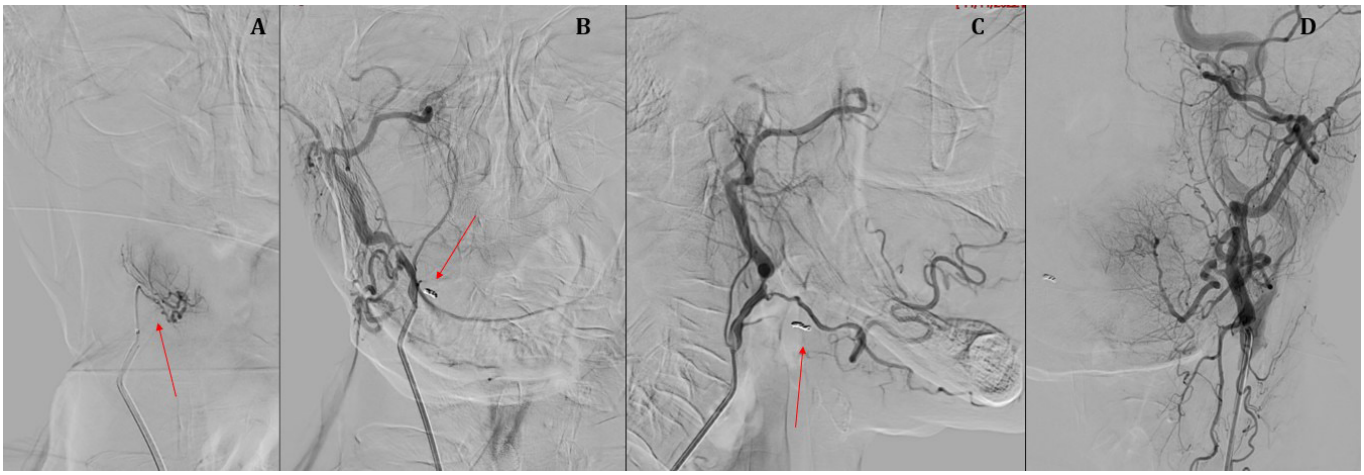

Fig. 3. Superselective arteriography of the right lingual artery (A); arteriography with diagnostic catheter, highlighting permanent embolic material (coil) in anteroposterior (B) and lateral (C) projections; Control angiography of the left common carotid artery, highlighting the vascularization of the left lingual artery after embolization of the right lingual artery (D)

quences affecting speech, swallowing, and loss of taste in the affected territory [10];

- Lingual Abscess: Secondary to ischemia and tissue necrosis, predisposing to infections that are difficult to treat in a compromised vascular territory. In severe cases, these can progress to sepsis [9];
- Recurrent Hemorrhage: In the context of an aggressive tumor infiltrating blood vessels and developing collateral circulation. The risk of recurrence is increased even after embolization [11];
- Systemic Complications: Including possible anaphylactic allergic reactions to the embolic material and the contrast agent used [12].

The main reasons for choosing coil embolization over other embolic agents are:

- Precision and Efficacy Control: Coils offer precise control, allowing specific blockage of the lingual artery or its branches, reducing blood flow without affecting adjacent areas. In the context of head and neck tumors, uncontrolled embolization can compromise healthy tissues and generate severe complications [13];
- Reduced Risk of Rebleeding: Compared to other embolic agents, permanent mechanical occlusion decreases the risk of vessel recanalization and rebleeding [14];
- Reduced Risk of Extensive Tissue Necrosis: Embolic agents, such as embolization particles, can disperse and occlude surrounding vessels, in this case, the anastomoses of the right lingual artery with the left one, increasing the risk of tongue necrosis [13];
- Prevention of Embolic Material Displacement: High blood flow and rich vascularization can increase the risk of displacement of embolic materials such as embolization particles. Therefore, coil embolization is more effective in high blood flow environments, ensuring the desired occlusion [15];
- Radiologic Marking: Visibility is essential in delicate and complex procedures in the head and neck area. Radiologic marking allows precise monitoring during the procedure and post-procedural control [15].

The coaxial system for superselective catheterization offers precision in interventions required in a restricted and specific vascular space. The advantage of the precision it offers reduces the risk of affecting adjacent healthy tissues. In managing bleeding at the base of tongue tumors, coil embolization provides precise and permanent occlusion.

COPD chronically affects lung function, and chronic liver disease can alter the metabolism and detoxification of medications, thus reducing the tolerability of oncological treatment.

Cumulative risk factors, such as smoking, chronic inflammation, and compromised general health, have favored the development and aggressive evolution of lingual neoplasm. Cachexia, combined with the inability to adequately feed and existing comorbidities, limits therapeutic options, complicating the prognosis and management of the disease.

Based on the physician's perception of the patient's condition and self-assessment, the ECOG-PS can be subjective but is useful for guiding therapeutic decisions or the patient's eligibility for certain treatments or palliative care. Progression to ECOG-PS 3 in a patient with lingual neoplasm results from a combination of factors such as local progression, associated symptoms (pain, dysphagia) and systemic effects of the disease (extreme fatigue, marked weight loss). The decline in performance can occur within a few months, depending on the specifics of the case. In conclusion, in a patient with an aggressive and advanced form of lingual neoplasm, progression to an ECOG-PS 3 score that affects autonomy is rapid [16].

Although endovascular intervention is often mentioned in the literature as being used for specific vascular injury post-radiochemotherapy, this case highlights the imperative need for therapy to allow the initiation of radiochemotherapy. Delaying the start of chemotherapy and using palliative irradiation in a patient presenting with hemorrhage from the lingual tumor is due to several factors con-

cerning patient safety and treatment efficacy. Hemorrhage control and prioritizing hemostasis are essential, given the fragility of tissues and the risk of recurrent bleeding. Due to the risks associated with chemotherapy, such as effects on coagulation and tissue healing, it is recommended to delay it until the patient's condition is stabilized. Post-embolization recovery and careful monitoring are necessary to ensure long-term bleeding cessation. After embolization, secondary complications may arise that require management before starting chemotherapy. Palliative irradiation can be an emergency or temporary measure for local tumor control, reducing its size, and controlling local symptoms. In urgent cases, where rapid symptom control is essential, radiotherapy may be preferred [17].

Endovascular embolization is an effective method for controlling severe bleeding in the head and neck region. The choice of technique and the planning of subsequent interventions are individualized based on the etiology of the hemorrhage and the specific characteristics of each patient.

The literature frequently highlights the use of embolization techniques in managing post-radiotherapy bleeding or carotid blowout syndrome. For instance, one case study describes a 45-year-old patient who developed carotid blowout syndrome following multiple therapies for laryngeal cancer, underscoring the importance of careful evaluation and a personalized management approach [18].

Compared to other cases described in the literature, embolization with permanent materials offers more precise hemostatic control than temporary agents, minimizing the risk of rehemorrhage, particularly in head and neck tumors with significant local invasion [19,20].

Similarities with other cases can be seen in the use of the coaxial system and microcatheters for increased selectivity. The literature describes embolizations performed for pseudoaneurysms, recurrent tumors, severe acute hemorrhages associated with inoperable neoplasms, lingual hemangiomas embolized prior to surgery, and severe hemorrhages from large facial arteriovenous malformations involving embolization of the branches of the internal maxillary artery [19-21].

These studies document embolizations of various arterial branches in the head and neck region. Additionally, some reports emphasize the necessity of repeat procedures, particularly in patients with a history of chemoradiotherapy [22].

Conclusion

The compromised general condition of the patient requires stabilization before initiating systemic chemotherapy. In this context of hemorrhage from the ulcerated lingual tumor and embolization of the source, delaying chemotherapy and using palliative radiotherapy can help stabilize the patient before starting aggressive systemic treatment, which may induce additional complications without providing significant long-term benefit. Due to the structural and functional impairment of the tongue and oropharyngeal cavity, patients with base of tongue cancer must be closely monitored for aspiration pneumonia [23].

Authors' contribution

DMH (Conceptualization; Visualization; Writing – original draft; Writing – review & editing)
MM (Data curation; Formal analysis; Validation; Investigation; Methodology; Supervision)
MR (Resources; Formal analysis; Investigation; Methodology; Supervision)
PC (Data curation; Formal analysis; Validation; Investigation; Methodology; Supervision)

Conflict of interest

None to declare.

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