

Lingual Artery Pseudoaneurysm after Total Laryngectomy: A Case Report

GAYATHRI LEKSHMI¹, SANTHI THANKAPPAN PILLAI², SUSAN JAMES³

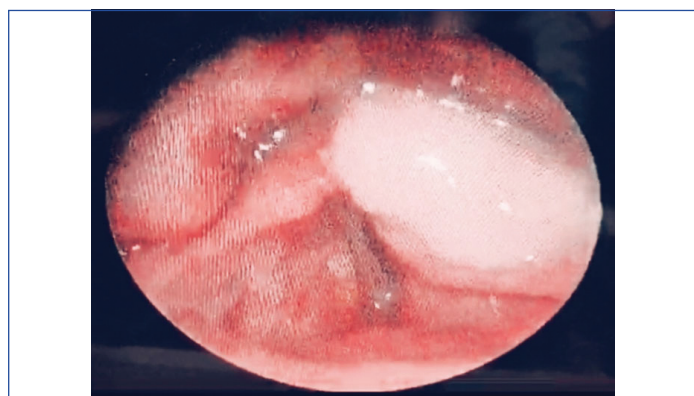
ABSTRACT

Pseudoaneurysm of the lingual artery is an extremely rare condition, and the most common causes are trauma, inflammation, neoplasm, or iatrogenic factors. An aneurysm refers to a weakening of an artery wall that creates a bulge or distention of the artery. Aneurysms of the external carotid artery and its branches are rare, comprising only 2.2% of cervical carotid aneurysms, with an even rarer occurrence of mycotic pseudoaneurysm. The superficial temporal artery and facial artery are the most commonly involved branches. Pseudoaneurysms of the lingual artery are an extremely rare entity and are often a consequence of neck surgery, trauma, inflammation, chemoradiotherapy, or odontogenic infection. They may cause life-threatening bleeding. Hereby, the authors present the case of a 47-year-old male patient with biopsy-proven Squamous Cell Carcinoma (SCC) of the hypopharynx who underwent total laryngectomy with Pectoralis Major Myocutaneous Flap (PMMC) reconstruction. The patient presented with a chief complaint of massive bleeding from the oral cavity three weeks post-procedure. The patient was immediately managed conservatively and started on inotropes. Computed Tomography (CT) angiogram of the neck showed a well-defined rounded enhancing structure in relation to the right lingual artery, with enhancement comparable to a vascular structure suggestive of a pseudoaneurysm. The patient underwent lingual artery embolisation, resulting in complete occlusion of the pseudoaneurysm. It is difficult to diagnose and manage lingual artery pseudoaneurysms, and they can cause severe distress to patients when bleeding occurs. To prevent morbidity and mortality associated with lingual artery aneurysms, they must be diagnosed and treated as early as possible.

Keywords: Angiogram, Embolisation, External carotid artery, Pectoralis major myocutaneous flap

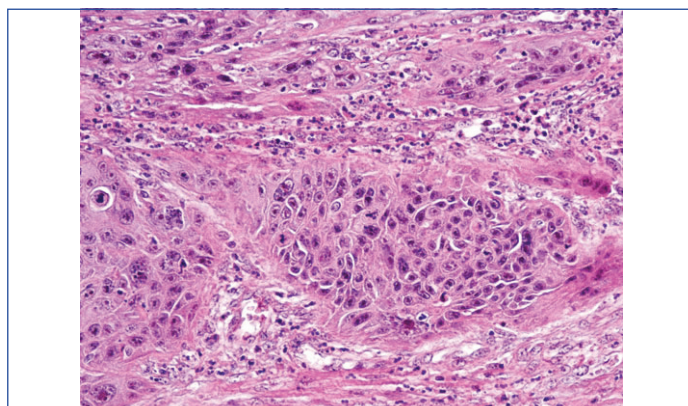
CASE REPORT

A 47-year-old male patient, who was a known case of carcinoma of the hard palate and treated with concurrent chemoradiation in 2017, presented in 2020 at the Outpatient department of Otorhinolaryngology with symptoms of hoarseness, difficulty in swallowing, foreign body sensation in the throat, and breathing difficulty for three months. Videolaryngoscopic examination revealed an ulceroproliferative growth involving the left pyriform sinus, left aryepiglottic fold crossing the midline with left hemilarynx fixity, and impaired mobility of the right vocal cord. No neck nodes were found [Table/Fig-1]. Magnetic Resonance Imaging (MRI) showed irregular, heterogeneously enhancing soft tissue thickening in the left half of the epiglottis, left pre-epiglottic space, bilateral aryepiglottic fold, left vocal cord, left vallecula, and left pyriform fossa, with thyroid cartilage erosion. Based on these findings, a provisional diagnosis of hypopharyngeal malignancy was made.



[Table/Fig-1]: Ulceroproliferative growth involving left pyriform sinus, left aryepiglottic fold and hemilarynx fixity.

A direct laryngoscopic biopsy from the lesion showed moderately differentiated SCC. The final diagnosis was carcinoma of the hypopharynx, T4aNOMO stage IVA {Tumour Node Metastasis (TNM) staging} [Table/Fig-2].



[Table/Fig-2]: Histopathological study showing moderately differentiated Squamous Cell Carcinoma (SCC).

The patient underwent total laryngectomy with partial pharyngectomy, bilateral modified radical neck dissection, and reconstruction with a PMMC flap [Table/Fig-3-7]. Postoperatively, the patient was monitored in the Intensive Care Unit (ICU). The vitals were stable, and the drain output was normal.

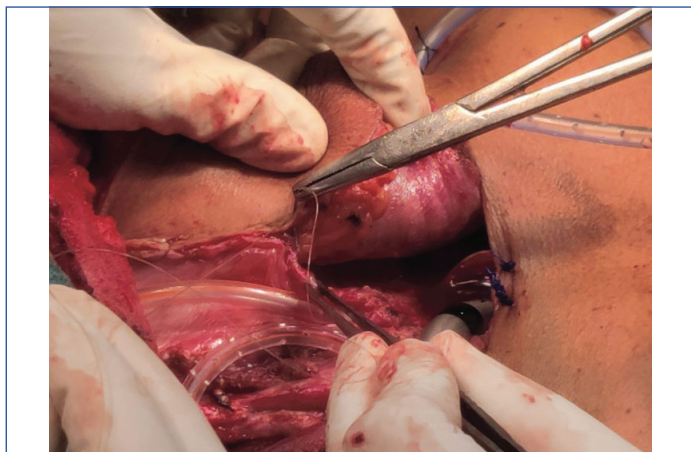


[Table/Fig-3]: Gluck Sorenson skin incision made.

[Table/Fig-4]: Subplatysmal flap raised. (Images from left to right)



[Table/Fig-5]: Malignant growth identified and tumour excised.
[Table/Fig-6]: PMMC flap elevation. (Images from left to right)



[Table/Fig-7]: Reconstruction of neopharynx with PMMC flap.

One week after the surgery, suture removal was performed. Two weeks postsurgery, minimal gaping and pus discharge were observed from the suture site, suspecting a pharyngocutaneous fistula. It was managed conservatively with Intravenous (i.v.) antibiotics (amoxicillin-clavulanate and metronidazole) and pressure dressing, which resulted in healing of the fistula. After three weeks, the patient experienced massive oral cavity bleeding with recurrent episodes. The patient was stabilised, and a CT angiogram was performed, revealing a contrast-filling outpouching measuring 7.5×6.3×7.2 mm arising from the mid-portion of the right lingual artery, suggestive of a saccular-type pseudoaneurysm [Table/Fig-8]. Digital Subtraction Angiography (DSA) showed the right lingual artery pseudoaneurysm, as shown in [Table/Fig-9].



[Table/Fig-8]: Computed Tomography angiogram shows pseudoaneurysm.
[Table/Fig-9]: Digital Subtraction Angiography (DSA) shows right lingual artery pseudoaneurysm. (Images from left to right)

Endovascular embolisation was performed using Polyvinyl Alcohol (PVA) and coils, which effectively controlled the bleeding. Subsequently, the patient did not experience any further bleeding. On regular follow-up, the patient has remained disease-free for the past two years.

DISCUSSION

Aneurysms are a result of the destruction or weakening of the arterial wall, leading to the distention of the artery. They manifest as a bulge in the artery [1]. Aneurysms commonly occur in the aorta and cerebral arteries. Aneurysms of the external carotid artery and its branches are rare and can be caused by various factors such as trauma,

infection, atherosclerosis, inflammation, surgery, or chemoradiation [2]. Depending on their shape, aneurysms can be classified as saccular, fusiform, cylindrical, dissecting, or pseudoaneurysms [3]. Pseudoaneurysms are focal, eccentric widenings of the lumen that extend beyond the vessel wall, with the absence of all layers of the arterial wall [4]. Lingual artery pseudoaneurysms are even rarer than pseudoaneurysms of the superficial branches of the external carotid artery (such as the superficial temporal artery and facial artery).

Pseudoaneurysms have also been reported at sites of arterial anastomosis following free flap reconstruction and surgical ligation [2]. Only a few cases of lingual artery aneurysms following total laryngectomy have been reported in the literature. A rare case of pseudoaneurysm of the lingual artery, secondary to concurrent intra-arterial chemotherapy via the superficial temporal artery for advanced tongue cancer, has also been reported [5].

Schechter reported that external carotid artery aneurysms accounted for 2.2% of all cervical carotid aneurysms, while lingual artery aneurysms were particularly rare, although their prevalence remains unclear [6]. An incidence of 0.11%, with only one case out of 926 oral cancer patients, has been reported [7]. While the majority of lingual artery pseudoaneurysms have a traumatic or surgical origin (especially tonsillectomy), some cases have been associated with radiofrequency [8], chemoradiation, infection, and malignancies of the head and neck, particularly in the oral cavity and oropharynx. Idiopathic aneurysms have also been described. In the literature, three cases of lingual artery pseudoaneurysms have been reported after total laryngopharyngectomy, where patients presented with haemorrhage two months after surgery, unlike the present case where the patient experienced oral cavity bleeding three weeks after surgery. Two cases of pseudoaneurysms were reported, one at the site of lingual artery ligation and the other at the site of arterial anastomosis after reconstruction with a jejunal graft, where the cases presented with haematemesis and a pulsating neck mass four weeks after surgery [9].

Aneurysms of the external carotid artery may present with oral cavity bleeding, swelling in the oral cavity, neck mass, or neurological complications. Contrast Enhanced CT (CECT) can be used to evaluate the post-treatment neck, especially in cases of suspected complications, as it is readily available. In the present case, CECT helped in identifying the subcentimetric vascular lesion. Contrast extravasation may be observed in cases of active haemorrhage. CT angiography is also useful for the diagnosis of vascular lesions. Digital Subtraction Angiography (DSA) is considered the gold standard for diagnosing cervical arterial vascular injuries. The commonly used treatment modalities for managing pseudoaneurysms include endovascular therapy (using coils, PVA particles, and n-butyl cyanoacrylate), ultrasound-guided percutaneous thrombin injection, and surgical resection [2]. In the present case, endovascular embolisation with PVA and coils was performed as the management approach.

CONCLUSION(S)

Pseudoaneurysms carry an inherent risk of rupture and can lead to massive, life-threatening bleeding. Therefore, it is crucial to promptly diagnose and treat pseudoaneurysms. Pseudoaneurysms should be considered in cases presenting with sudden oral bleeding following neck surgeries, intra-arterial chemotherapy, or neck irradiation for advanced head and neck cancer. Early identification and treatment of these complications can significantly reduce mortality rates.

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PLAGIARISM CHECKING METHODS: [Jain H et al.]

- Plagiarism X-checker: Jun 14, 2023
- Manual Googling: Sep 23, 2023
- iThenticate Software: Oct 14, 2023 (20%)

ETYMOLOGY: Author Origin

EMENDATIONS: 5

AUTHOR DECLARATION:

- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

Date of Submission: Jun 12, 2023

Date of Peer Review: Sep 05, 2023

Date of Acceptance: Oct 22, 2023

Date of Publishing: Jan 01, 2024