

Carotid blowout syndrome from laryngeal chondroradionecrosis requiring extra-anatomical vascular bypass: a case series

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Introduction

Laryngeal chondroradionecrosis (CRN) is an uncommon late-stage complication of radiation therapy for head and neck cancer during which local tissue hypoxia leads to inflammation and ultimately cell death.¹ Carotid blowout syndrome (CBS) is another potential complication of radiotherapy and can be life-threatening.^{2,3} Both have been well documented; however, there is a paucity of literature describing the relationship between the two and the management of this challenging process. This study describes three cases of patients who developed CBS secondary to pseudoaneurysms from laryngeal CRN and underwent successful treatment with total laryngectomy and extra-anatomical vascular bypass.

Case 1

A 36 year old female diagnosed with T2 N0 M0 squamous cell carcinoma (SCC) of the glottis in January 1995 completed primary hyperfractionated radiotherapy in March 1995. In May 2007, she developed sudden-onset projective hematemesis secondary to a right carotid-pharyngeal fistula. An endovascular stent was placed in the right internal carotid artery (ICA); however, she experienced two further carotid blowouts within the next 2 weeks, both also requiring endovascular stenting. This resulted in left hemiparesis from a thromboembolic infarct. In June 2007, surgical exploration revealed extensive CRN with perforation of the pharynx and piriform sinus on the left and frank purulence along the superior horn of the right thyroid cartilage. She had a perforated right carotid artery with 4 cm of exposed stent visualized in her pharynx. She underwent a total laryngectomy, near total pharyngectomy, pharyngeal reconstruction with a pectoralis major myocutaneous flap (PMMF), resection of the carotid artery, and right carotid-to-carotid bypass with a greater saphenous vein graft. On post-operative day (POD) #9, she suffered an acute chest wall bleed. A CT Angiogram of the neo-carotid artery revealed a pseudoaneurysm. She then underwent superinfected graft excision, chest wall hematoma evacuation, and a right axillary artery to right ICA extra-anatomical bypass using a translocated saphenous vein graft. She ultimately recovered completely and was discharged. She developed a postoperative pharyngeal leak that resolved with conservative management with placement of a salivary bypass tube. Since that time, she has had no further complications and her vascular bypass has remained patent.

Case 2

A 63 year old male diagnosed with T2 N0 M0 SCC of the supraglottis in February 2009 completed primary radiotherapy in May 2009. In August 2009, due to concern for tumor recurrence on routine surveillance, direct laryngoscopy was performed revealing an ulcerative lesion of the vallecula involving the left and right pharynx, left epiglottis, and subglottic larynx. There was exposed thyroid cartilage and hyoid bone in the pharynx bilaterally. Pathology revealed highly advanced chondroradionecrosis with minimally invasive SCC in the subglottis. In November 2009, he had large volume carotid blowout. Angiography demonstrated a 4 mm pseudoaneurysm and erosion of the left external carotid artery (ECA) at the takeoff from the common carotid artery (CCA). He was not a candidate for endovascular intervention, so he underwent a left axillary artery to left ICA extra-anatomical bypass with a greater saphenous vein graft. Days later, he was taken for total laryngectomy and near total pharyngectomy with reconstruction with a PMFF. He developed a postoperative 3.0 x 3.0 cm chest wall hematoma which did not require surgical intervention. He developed no further complications.

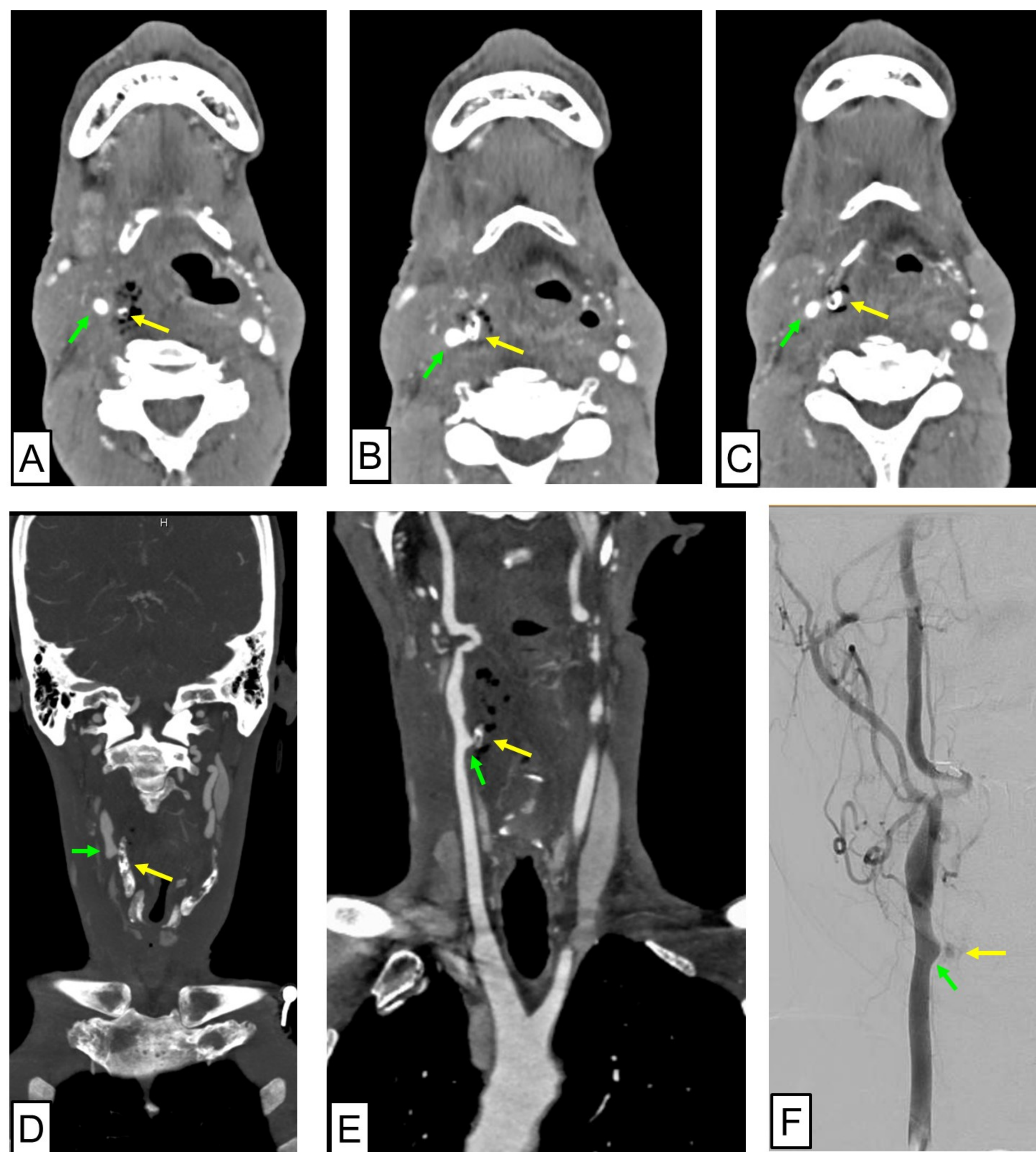


Figure 2: Radiographic findings in chondroradionecrosis and carotid blowout. **A-C)** CT Angiogram, axial view. The superior horn of the right thyroid cartilage has surrounding soft tissue edema and emphysema (yellow arrow). Note the proximity of the right common carotid artery (green arrow). **D-E)** CT Angiogram, coronal view. The superior horn of the right thyroid cartilage has a fragmented appearance (yellow arrow). Note the pseudoaneurysm of the common carotid artery in contact with the cartilage (green arrow). **F)** Angiography of the right common carotid system. Note the pseudoaneurysm of the common carotid artery (green arrow) just proximal to the bifurcation and the extravasation of contrast medially, representing active bleeding (yellow arrow).

Table 1: Summary of case characteristics

	Case 1	Case 2	Case 3
Age at diagnosis (years)	36	63	57
Cancer histology	Squamous cell carcinoma	Squamous cell carcinoma	Squamous cell carcinoma
Cancer location	Glottis	Supraglottis	Oropharynx
TNM Staging	T2 N0 M0	T2 N0 M0	T3 N2 M0
Prognostic Staging	II	II	II
Treatment modality	Primary radiotherapy	Primary radiotherapy	Chemoradiation
Radiation dose	Unknown	68 Gy	70 Gy
Time from treatment to carotid blowout (months)	145.3	6.1	10.3
Location of pseudoaneurysm	Neo-carotid artery (venous graft)	External carotid artery	Common carotid artery
Endovascular intervention	Yes (x 3)	No	Yes (x1)
Neurologic sequelae from endovascular intervention	MCA, ACA infarct with hemiparesis	N/A	No
Location of radionecrosis	Superior horn of thyroid cartilage	Superior horn of thyroid cartilage, hyoid bone	Superior horn of thyroid cartilage, hyoid bone
Malignancy Present	No	Yes (subglottis)	No
Surgery performed	TL, partial pharyngectomy, PMFF	TL, partial pharyngectomy, PMFF	TL, partial pharyngectomy, PMFF
Extra-anatomical bypass performed	Axillary to internal carotid	Axillary to internal carotid	Axillary to internal carotid
Post-operative complications	Chest wall hematoma requiring operative evacuation; pharyngeal leak resolved with salivary bypass tube	Chest wall hematoma, did not require intervention	Chest wall & neck hematoma requiring operative evacuation and primary repair of anastomosis
Survival since last CBS event (months)	190.3	161.2	5.7

Case 3

A 57 year old male diagnosed with a T3 N2 M0 p16(+) SCC of the right tonsil in August 2021 completed chemoradiation in December 2021. On routine surveillance there was concern for tumor recurrence, so he underwent direct laryngoscopy and biopsy which were negative for malignancy but demonstrated cellular necrosis on two separate occasions. In October 2022, he experienced a large volume oropharyngeal bleed that ceased spontaneously. Emergency surgery revealed a large necrotic pharyngeal ulcer with exposed hyoid bone. Postoperatively, he experienced further large volume hematemesis requiring emergent endovascular intervention. Angiography revealed a defect in the right common carotid artery at the level of the superior horn of the thyroid with an associated large right carotid pseudoaneurysm which was stented and stabilized. Consequently, he sustained a small asymptomatic distal MCA thromboembolus. Ten days later, he underwent a total laryngectomy, left PMFF reconstruction, and extra-anatomical carotid artery bypass. Intraoperatively, he had a third oropharyngeal carotid blowout controlled with oral gauze packing and a temporary vascular shunt from the CCA to the ICA. Once stabilized, this was converted to a right axillary to ICA extra-anatomical bypass. On POD #1, he developed a leak in the graft anastomosis with the ICA that resulted in a hematoma and required evacuation and primary repair of the distal anastomosis. He had no additional complications, neurological sequelae, or disease recurrence.

Discussion

Laryngeal chondroradionecrosis is a relatively uncommon late-stage complication of radiotherapy, with incidence ranging from <1% to 10.7%.⁴⁻⁶ This can be a devastating complication of radiotherapy to the head and neck, as up to 25% of patients ultimately require a laryngectomy due to aspiration and organ dysfunction.¹ Due to the proximity of the carotid arterial system with the laryngeal framework, infection in this area may predispose the arterial walls to develop a pseudoaneurysm with resultant weakening and subsequent rupture.⁵ In this series, all three patients had laryngeal CRN resulting in cartilage exposure within the pharynx and ensuing inoculation with aerodigestive bacteria. This infection then precipitated a pseudoaneurysm in the adjacent artery ultimately leading to CBS.

Current methods of managing CBS include endovascular intervention, open arterial ligation, or vascular bypass.² Endovascular intervention, including endovascular embolization or endovascular stent placement, can be performed quickly in emergent situations and may allow for the control of hemorrhage. However, it risks neurologic morbidity and high rates of rebleeding and stent occlusion.^{2,3,7} Both patients in this series who underwent endovascular stenting developed cerebral thromboemboli and rebleeding. For these reasons, vascular bypass may be a preferred long-term solution for patients with a longer expected survival period.⁷

At the time of this publication, all three patients in this series are currently living and maintain patency of their extra-anatomical anastomoses. There are several factors contributing to this positive outcome. First, the extra-anatomical bypass re-routes arterial flow away from the infected wound while a total laryngectomy removes the source of infection and thus the underlying cause of the pseudoaneurysm. This was well illustrated in Patient 1, where the first attempt at vascular bypass included placement of a graft within a wound bed with residual infection. This resulted in a pseudoaneurysm of that graft and subsequent rebleeding. It was only definitively managed after an extra-anatomical bypass. Secondly, sacrifice of the larynx was an acceptable consequence when weighed against the risk of future rebleeding episodes, which portends neurologic morbidity or mortality in up to 50% and 60% of patients, respectively.² Third, CBS in these cases was associated with infection rather than malignancy. Finally, patients in this study all had early stage head and neck cancer. CBS patients with advanced state of tumor disease have worse medium- and long-term survival than their early stage counterparts.⁷

Conclusion

Laryngeal chondroradionecrosis is an uncommon but potentially devastating long-term complication of radiotherapy for head and neck cancer due to its potential to cause carotid blowout syndrome. In these patients, a total laryngectomy with extra-anatomic vascular bypass has been effective in eliminating the causative infection while establishing long-term patent arterial flow away from diseased tissue.

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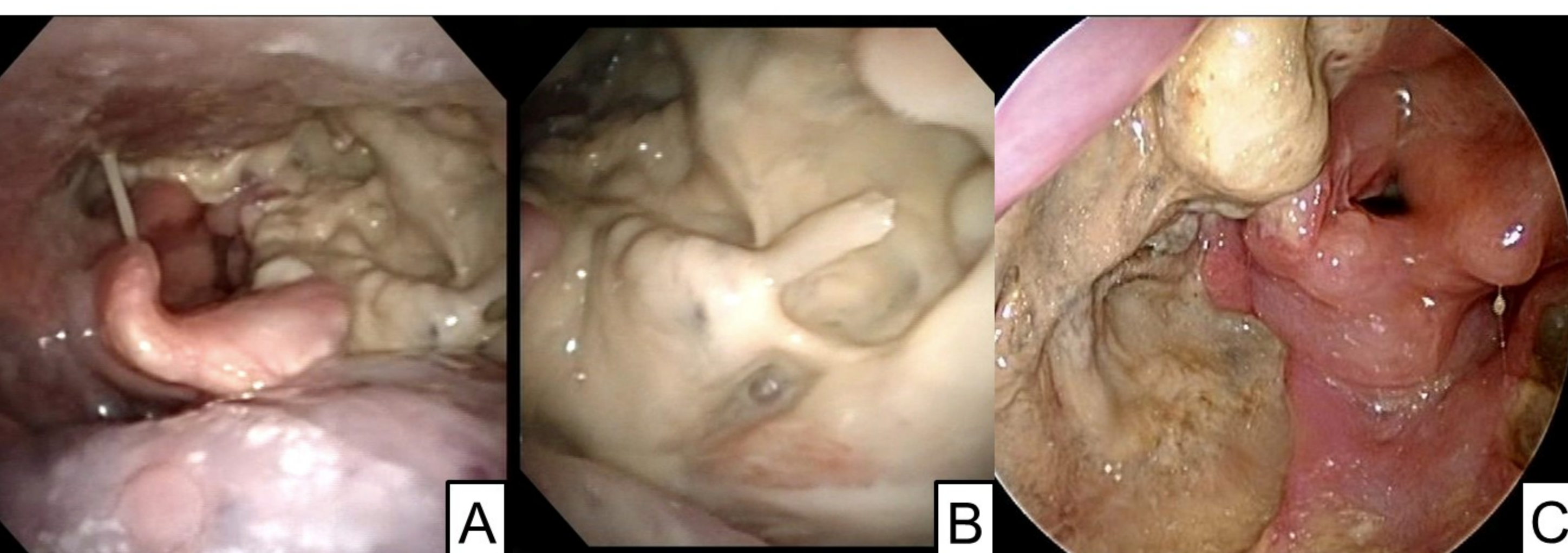


Figure 1: Chondroradionecrosis findings on endoscopy. **A)** Nasolaryngoscopy with necrotic changes of left vallecula, pharyngeal walls, epiglottis, and aryepiglottic fold. **B)** A magnified view of the left hypopharynx on nasolaryngoscopy. Note the exposed and necrotic hyoid bone. **C)** Operative findings during direct laryngoscopy. Note the exposed thyroid cartilage. Arterial pulsations were noted just lateral to the necrotic area of the left hypopharyngeal wall.

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