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Osteoradionecrosis of the hyoid bone complicated by pharyngocutaneous fistula: A case report and literature review

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ABSTRACT

Background: Osteoradionecrosis (ORN) is a well-known complication following irradiation of head and neck malignancies. ORN commonly occurs in the mandible but is rarely reported in the hyoid bone.
Case presentation: A 76-year-old female with a history of oropharyngeal squamous cell carcinoma presented with pharyngocutaneous fistula 14 years after primary chemoradiation. Imaging showed necrosis of the hyoid bone. She underwent excision of the hyoid to rule out malignancy. Pathology was negative for carcinoma, but did show extensive fragmentation and bony necrosis consistent with ORN. The patient's clinical course, surgical treatment, and management considerations are discussed here.
Conclusions: Hyoid ORN should remain in the differential during diagnostic workup of previously irradiated head and neck cancer patients. The presentation of a pharyngocutaneous fistula should prompt workup to rule out malignancy before assigning a diagnosis of ORN.

1. Introduction

Osteoradionecrosis (ORN) is a well-known complication following irradiation of head and neck malignancies. ORN commonly occurs in the mandible but is rarely reported in the hyoid bone. While there has been increased awareness of hyoid ORN, we present the first documented case of ORN involving the hyoid complicated by a pharyngocutaneous fistula.

2. Case presentation

A 63-year-old female presented with severe oral pain, weight loss, and dysphagia. She was found to have a left base of tongue mass, which was diagnosed as squamous cell carcinoma in 2004. Her medical history included rheumatoid arthritis, glaucoma, hypertension, mild coronary artery disease, and borderline diabetes. Despite a lifelong history of heavy smoking, the patient had abstained from tobacco and alcohol for over a decade at the time of presentation.

She underwent a course of concurrent chemoradiation to treat her oropharyngeal cancer, including 3 cycles of 100 mg/m^2 cisplatin and 70 Gy of external beam radiation delivered to areas of gross involvement. She remained free of disease following treatment, but 12 years later, presented with pain and swelling of the jaw. Intra-oral examination revealed areas of exposed mandibular bone. Computed tomography (CT) imaging showed evidence of bony necrosis in the right mandible. This was determined to be early-stage ORN and she subsequently underwent debridement of the mandible and advancement of mucosal flaps to provide coverage of the exposed bone.

Over the following year, the patient's lower jaw pain worsened. Panorex revealed progression of ORN. She subsequently underwent a right segmental mandibulectomy and reconstruction with a fibular free flap. Her early post-operative course was uneventful. However, two

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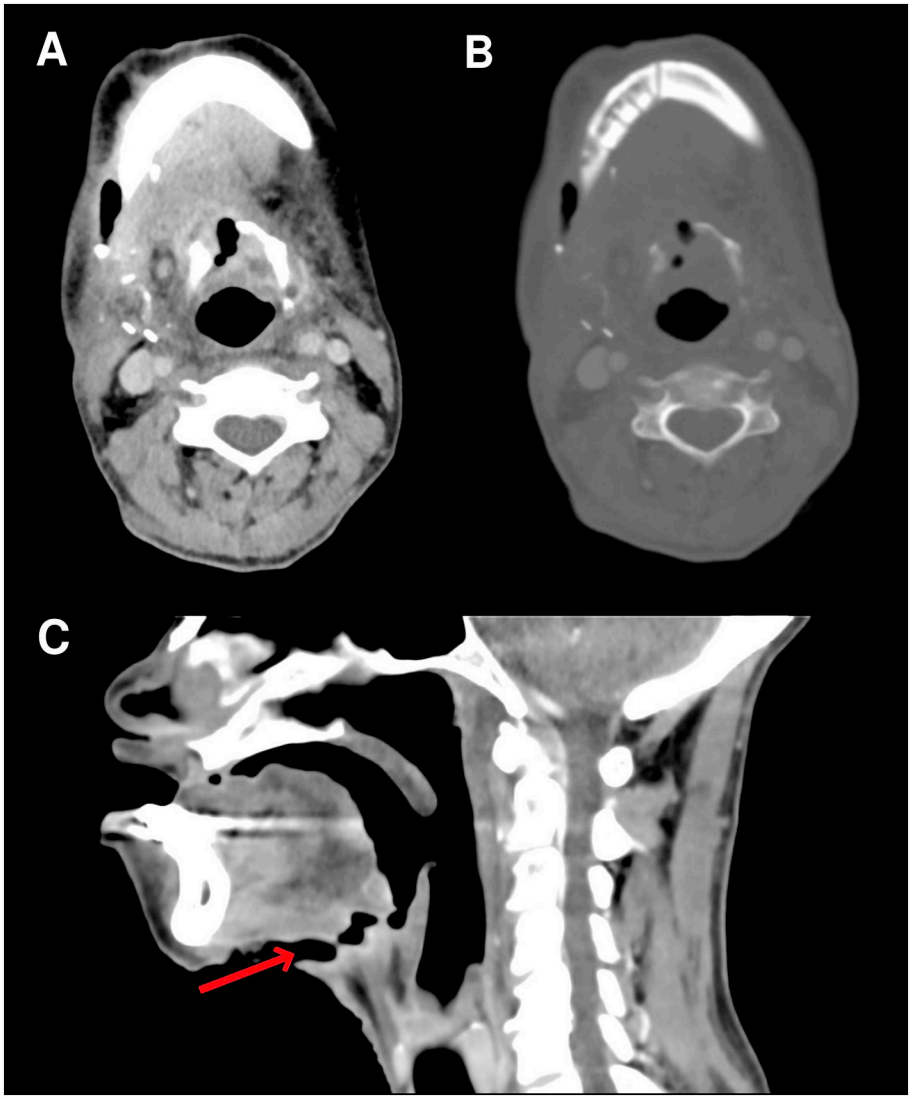

Fig. 1. Contrast CT images demonstrating hyoid fragmentation and adjacent air pockets in A) soft tissue and B) bone windows. C) Fistula tract (arrow) from the submental region to the valeculla is noted on sagittal images.

months later, she developed a spontaneous submental wound dehiscence. A panorex revealed good union between the fibula and the native mandible. On examination, part of the mandibular hardware was visible, which was thought to be the etiology of her wound breakdown. The mandibular hardware was removed.

Negative pressure therapy was used to help expedite wound healing but no improvement of her wound was evident. On closer examination, a fistula tract from the submental wound to the valeculla was detected. With liquid intake, there was obvious leakage from the pharynx through the fistula tract. A CT scan was obtained, demonstrating fragmentation and bony demineralization of the hyoid with air pockets in the adjacent soft tissues (Fig. 1). An associated air pocket at the level of the hyoid defect with fistulization to the submental region was also seen.

The patient was then taken to the operating room for biopsy to rule out malignancy. Frozen section of adjacent soft tissue was negative for malignancy. The hyoid bone was then exposed bilaterally revealing clear devitalization (Fig. 2). The suprahyroid and infrahyoid musculature was released quite easily from the hyoid, which was then removed and the wound was gently packed. Final pathology demonstrated chronic osteomyelitis and radiation-induced changes within the hyoid bone specimen, which was consistent with ORN.

3. Discussion

ORN has been a common finding associated with radiation treatment of head and neck malignancies [1]. The incidence of ORN ranges from 2% to 10% of patients who undergo radiotherapy for head and neck cancers [2,3]. The median latency period between irradiation and ORN detection is 1–2 years [3]. The diagnosis of ORN requires that recurrent or persistent neoplastic disease be ruled out. ORN is commonly associated with exposed bone, pain, and radiographic evidence of osteolysis.

While the diagnosis of mandibular ORN is fairly uncomplicated,

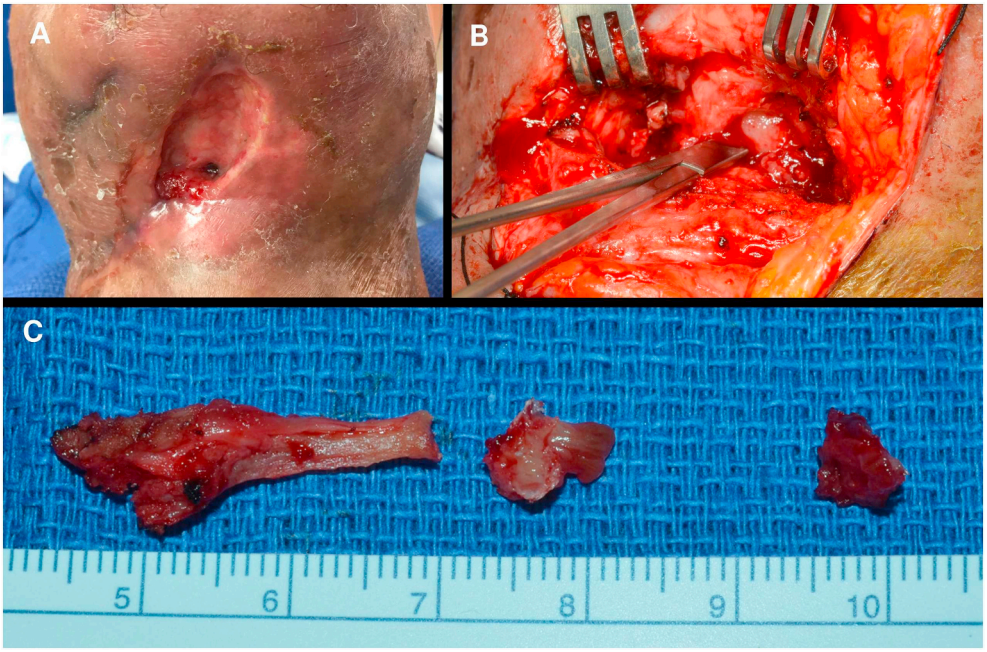

Fig. 2. A) Pre-operative photograph showing open wound of the anterior neck with an associated pharyngocutaneous fistula. B) Intraoperative photograph demonstrating a surgical clamp inserted into the fistula terminating in the pharynx. C) Hyoid resection specimen demonstrates areas of fragmentation and necrosis.

ORN of the hyoid bone may be under-recognized. Previously reported cases are listed in Table 1. Retrospective imaging reviews have demonstrated the presence of hyoid ORN at a higher rate than might be expected by clinicians. Hatakeyama et al. reviewed imaging of 39 previously irradiated oropharyngeal cancer patients, finding that 12 patients showed evidence of ORN in the hyoid on magnetic resonance imaging (MRI) (Table 1) [4]. However, only two of these 12 patients had associated symptoms, which commonly included new onset dysphagia and odynophagia, and occasionally non-specific neck pain [4–7]. In their study of 13 patients with imaging findings highly suggestive of hyoid ORN, Yoo et al. report that eight patients (61.5%) presented with new onset dysphagia, with or without odynophagia [8]. Thus, clinical diagnosis of ORN of the hyoid may be difficult given the non-specific nature of symptoms, particularly in patients who have undergone prior surgical and radiation treatment.

Pharyngocutaneous fistula related to ORN of the hyoid has not been previously reported. Monceaux et al. reported in 1999 a case of hyoid ORN which was associated with purulent drainage to the lateral neck, but no violation of the pharynx was encountered [9]. Hatakeyama et al. also reported that one case from their series demonstrated erosion from the hyoid into the pharynx, but no fistulization to the skin was present [4]. Spontaneous pharyngocutaneous fistula can arise from infection, trauma, the presence of a foreign body, or malignancy. Particularly in patients with a prior history of cancer, the development of a fistula should raise suspicion of recurrence and prompt workup to rule out malignancy. Adequate treatment of pharyngocutaneous fistulas relies on addressing the underlying cause. In our case, removal of necrotic bone was necessary to clear the associated infection and expedite granulation of the wound bed. Because patients with ORN have decreased tissue healing as a result of radiation treatment, regional or free tissue transfer may be required for definitive repair. In our case, a pectoralis major flap was required to achieve closure.

Radiologic findings of hyoid ORN on CT imaging include hyoid fragmentation or cortical disruption, intraosseous air, and the absence of soft tissue enhancement to suggest associated malignancy [8]. On MRI, the fibrous changes associated with ORN are indicated by partial or total low signal on both T1 weighted images (T1WI) and T2 weighted images (T2WI), respectively, while normal intact hyoid bone is represented by high signal on T1WI in bone marrow and decreased signal on T2WI [4]. A review of the literature regarding the findings on PET imaging of hyoid ORN is limited. Yoo et al. described four patients with hyoid ORN who underwent PET scans, three of which showed focal avidity with standard uptake values (SUVs) between 6.8 and 7.5; one patient did not demonstrate any uptake [8]. Under-recognition of hyoid ORN may also be attributable to the relatively small volume of the hyoid compared to the mandible, making detection less likely on imaging [4]. The detection of hyoid ORN also may be limited if fine-cut scans are not performed. Surgical removal of the bone is the most common approach to manage hyoid ORN. Given the burden to rule out malignancy, biopsy should be obtained to guide treatment and management discussions. If left untreated, ORN of the hyoid can lead to severe neck infection, laryngeal stenosis, and even carotid rupture [4,6,7].

4. Conclusion

We report the rare finding of osteoradionecrosis (ORN) of the hyoid bone and pharyngocutaneous fistula appearing 14 years after chemoradiation to treat squamous cell carcinoma in the tongue base. In order to prevent infection and other complications, clinicians should consider hyoid ORN during diagnostic workup of previously irradiated head and neck cancer patients who present with new symptoms and associated soft tissue changes.

Table 1
Review of previously reported cases of osteoradionecrosis in the hyoid. Asterisk (*) denotes re-irradiation.

Author, year (case #)	Age, sex	Primary tumor site	Staging	RT dose	RT type	Time between RT and ORN detection (months)	Hyoid ORN treatment	Disease-free follow-up (months)
Bhatia et al., 1979	55 M	Pyriform fossa	T?N3	60 Gy	Cobalt-60	4	No intervention	9
Echavez et al., 1992	66 M	Supraglottic	T4N2aMX	72 Gy	Electron beam	48	Local wound care and antibiotics	Unknown
Robertson et al., 1995	58 M	Laryngeal surface of epiglottis	T3N2aMX	80 Gy	Electron beam	4	Surgical resection	Unknown
Monceaux et al., 1999	80 F	Tongue base	T3N0M0	66 Gy	Electron beam	6	Surgical resection	6
Monceaux et al., 2nd case	60 M	Vallecular, epiglottis, tongue base	T4N0M0	70 Gy	Electron beam	5	Surgical resection	27
Monceaux et al., 3rd case	37 M	Tongue base	T3N0M0	70 Gy	Cobalt-60	180	Surgical resection	5
Smith et al., 2003	56 M	Tongue base	T2N2cM0	53 Gy	Photon beam	8	Endoscopic removal	Died post-op
Chard et al., 2012	54 M	Epiglottis	T2 N0	72, 54 Gy*	External beam	32	Surgical resection	Died of esophageal cancer 4 months later
Chard et al., 2nd case	59 F	Initial tumor in pyriform sinus; recurrences in oral cavity, tongue base	Unknown	Unknown*	Unknown	15 years	Surgical resection	Died of recurrent disease 15 months later
Yoo et al., 2010	67 M	Tongue base, n = 6	Not specified	Unknown	Not specified	34	1 laryngectomy	12
Yoo et al., 2nd case	46 M	Tonsil, n = 2		71 Gy		5	1 hyperbaric oxygen	6
Yoo et al., 3rd case	66 M	Supraglottic larynx, n = 2		60, 50.2 Gy*		2	therapy	23
Yoo et al., 4th case	64 M	Lateral pharyngeal wall not limited to		70 Gy		5	11 not treated	30
Yoo et al., 5th case	45 M	tonsil, n = 3		66.8 Gy		5.5		1
Yoo et al., 6th case	64 M			70.6 Gy		13		16
Yoo et al., 7th case	54 M			70.1 Gy		8		7
Yoo et al., 8th case	49 M			71.6 Gy		26		16
Yoo et al., 9th case	70 M			60, 20 Gy*		6		14
Yoo et al., 10th case	67 M			Unknown		1		21
Yoo et al., 11th case	47 M			Unknown		32		7
Yoo et al., 12th case	67 M			41, 63 Gy*		10		2
Yoo et al., 13th case	61 M			50, 60 Gy*		17		4
Hatakeyama et al., 2017 (12 cases in aggregate)	Median age: 60 79% M	Oropharyngeal (anterior wall n = 34, lateral wall n = 5)	85% Stage IV disease	65–70 Gy	Not specified	Median: 6	2 surgical resections 10 not treated	Unknown

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Declarations of interest

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References

[1] Murray CG, Herson J, Daly TE, Zimmerman S. Radiation necrosis of the mandible: a 10 year study. Part II. Dental factors; onset, duration and management of necrosis. Int J Radiat Oncol Biol Phys 1980;6:549–53.

[2] Nabil S, Samman N. Risk factors for osteoradionecrosis after head and neck radiation: a systematic review. Oral Surg Oral Med Oral Pathol Oral Radiol 2012;113:54–69. https://doi.org/10.1016/j.tripleo.2011.07.042.

[3] Mendenhall WM, Suárez C, Genden EM, de Bree R, Strojan P, Langendijk JA, et al. Parameters associated with mandibular osteoradionecrosis. Am J Clin Oncol 2018. https://doi.org/10.1097/COC.0000000000000424.

[4] Hatakeyama H, Fujima N, Tsuchiya K, Mizoguchi K, Mizumachi T, Sakashita T, et al. Osteoradionecrosis of the hyoid bone after intra-arterial chemoradiotherapy for oropharyngeal cancer: MR imaging findings. Cancer Imaging Off Publ Int Cancer Imaging Soc 2017;17:22. https://doi.org/10.1186/s40644-017-0123-7.

[5] Bhatia PL, Dutta NK, Sanasam JC. Osteonecrosis of hyoid bone and thyroid cartilage. Arch Otolaryngol 1979;105:553–4.

[6] Chard R, Monroe MM, Andersen PE. Hyoid osteoradionecrosis associated with carotid rupture: report of 2 cases. Head Neck 2013;35:E36–8. https://doi.org/10.1002/hed.21847.

[7] Curi MM, Cardoso CL, de Lima HG, Kowalski LP, Martins MD. Histopathologic and histomorphometric analysis of irradiation injury in bone and the surrounding soft tissues of the jaws. J Oral Maxillofac Surg Off J Am Assoc Oral Maxillofac Surg 2016;74:190–9. https://doi.org/10.1016/j.joms.2015.07.009.

[8] Yoo JS, Rosenthal DI, Mitchell K, Ginsberg LE. Osteoradionecrosis of the hyoid bone: imaging findings. AJNR Am J Neuroradiol 2010;31:761–6. https://doi.org/10.3174/ajnr.A1892.

[9] Monceaux G, Périé S, Montravers F, Angelard B, Corlieu P, St Guily JL. Osteoradionecrosis of the hyoid bone: a report of 3 cases. Am J Otolaryngol 1999;20:400–4.