CASE REPORT

Osteoradionecrosis of the hyoid bone – a novel application of the Sonopet ultrasonic aspirator

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SUMMARY

A 66-year-old man with a history of squamous cell carcinoma of the right tonsil presented to the emergency department with two life-threatening episodes of haemoptysis 9 months after completion of chemoradiation. He was evaluated to rule out recurrent malignancy, and was determined to have osteoradionecrosis of his hyoid bone that had led to an oropharyngeal fistula. Given the proximity to branches of the external carotid, he was offered tracheostomy and resection of the protruding necrotic bone with ultrasonic bone aspirator, which was placed down the shaft of the laryngoscope to minimise damage to adjacent structures. He tolerated the procedure well with complete resolution of exposed bone on follow-up laryngoscopy.

BACKGROUND

Osteoradionecrosis (ORN) is a non-healing area of devitalised bone in a radiated field that persists for at least 3 months.¹ The overlying skin or mucosa may or may not be intact.² It is an important and serious complication of radiation therapy (RT) performed for the treatment for head and neck cancers. ORN most commonly involves the mandible (80%).³ Other areas of the head and neck can be affected as well, including the maxilla, temporal bone, skull base, larynx and, least commonly, the hyoid bone. It is important to consider ORN in the differential when there is suspicion of a neoplastic recurrence in a patient previously treated with RT; ORN can similarly present as a mass with pain and soft tissue or bony destruction.

Symptomatic ORN with fracture may require surgical resection. Significant risks exist with traditional open resection, which can potentially worsen the patient's condition; inflamed, irradiated tissue and bone are unpredictable and slow to heal.² Our patient was successfully treated with a transoral endoscopic ultrasonic resection of the hyoid bone, which minimises collateral damage to surrounding structures.^{4–7}

In our review of the English otolaryngology literature, there have only been six documented patient cases of ORN involving the hyoid bone.^{8–11} An additional 13 patients have been described in the radiology literature.¹² To the best of our knowledge, only one previous case report has documented hyoid ORN presenting with haemoptysis,⁹ whereas no report has documented management of hyoid ORN with ultrasonic technology.

CASE PRESENTATION

A 66-year-old man with advanced squamous cell carcinoma of the right tonsil was treated with induction chemotherapy for 9 weeks, which was followed by 72 Gy of concurrent cisplatin-based chemoradiation. Nine months after completing chemoradiation, the patient experienced two episodes of severe and life-threatening haemoptysis. He was hospitalised for dehydration and anaemia. On physical examination he was found to be bleeding from the superior tonsillar fossa on the right side; he was taken to the operating room at an outside hospital for cauterisation of his tonsillar bleed. At the time, no abnormalities were noted aside from the presence of pseudomembrane and necrosis in the mid and inferior aspect of the tonsillar fossa. Two months after his hospitalisation, the patient presented to our clinic with right neck pain posterior to the mandible, at the site of the primary tumour.

Flexible fiberoptic laryngoscopy revealed pseudomembrane and erosion of the mucosa overlying the greater horn of the hyoid bone in the vallecula. The greater horn of the hyoid was noted to be protruding intact into the lumen of the oropharynx. The lesion moved with external digital manipulation of the patient's hyoid bone, confirming the identity of the bone internally. Neither active bleeding nor an obvious recurrent tumour, which is most likely to present as a discrete mass, was noted (figure 1).

A CT angiogram was unable to reveal the source of the significant bleed. The patient continued to experience pain, especially when turning his head to the contralateral side. Although the patient did not have a recurrent episode of haemoptysis, he did occasionally taste blood in his oral cavity. On presentation, the patient had been unable to

Figure 1 Endoscopic view of the supraglottic region. The arrow points to the eroded hyoid bone into the pharynx.

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maintain adequate caloric intake due to trismus and pain. He was placed on tube feeds while inpatient.

DIFFERENTIAL DIAGNOSIS

Deemed more appropriate to be included in discussion.

OUTCOME AND FOLLOW-UP

The benefits and risks of surgery were discussed with the patient. Owing primarily to the proximity of the external carotid artery and its branches to the lateral aspect of the hyoid bone, it was determined that hyoid bone debridement with tracheostomy for airway management would be the best method for removing the remnant of exposed hyoid bone in the pharynx. Accordingly, the Steiner expanding pharyngoscope was introduced transorally. Exposure of the oropharynx and Valledupar was extremely difficult due to narrow oral aperture and radiation-induced fibrosis. The patient was then suspended from the Mayo stand. Standard bone cutting instruments could not be introduced through the Steiner scope, as the aperture could not be expanded. The surgical site was noted to be quite inflamed and the tissue was quite friable. Thick endolaryngeal mucus was noted, which was consistent with the patient's history of RT. A Sonopet ultrasonic bone aspirator with an extended tip was utilised to ultrasonically resect the hyoid bone down to a base of bleeding bone under the pharyngeal mucosa. Two simple sutures of 4–0 Vicryl were placed in an attempt to bring mucosal edges together using an endoscopic technique.

The patient tolerated the procedure well; he was noted to have significant oropharyngeal mucus on his first postoperative visit, but without any exposed bone (figure 2). He was successfully decannulated several weeks later.

DISCUSSION

Pathogenesis

The theory behind the pathogenesis of ORN has witnessed several revisions since Meyer¹³ first posited that trauma to the weakened and irradiated bone made it susceptible to bacterial infection in 1970. In 1983, Marx¹⁴ discounted this theory, and argued that hypoxia drove the pathogenesis of ORN. He argued that RT induced endarteritis and microthrombi, leading to a lack of oxygen and energy in the bone and surrounding soft tissue. The resultant hypoxia caused destruction of osteoblasts and osteoclasts, potentiating bone and tissue necrosis while

preventing repair and wound healing^{14 15}; today, this theory is known as the hypoxic-hypovascular-hypocellular theory.

Recently, in the fibrotic-atrophy theory, Delanian and Lefaix¹⁶ posited that radiation-induced dysregulation of fibroblast activity results in an imbalance between the synthesis and degradation of tissue, leading to increased tissue atrophy and fibrosis within the damaged area. RT damages vascular endothelium, which causes these cells to release cytokines and growth factors that stimulate unregulated fibroblast activity, producing an extracellular matrix that is increasingly disorganised and fibrosed.¹⁵ This results in hypocellular bone, which, in combination with microthrombi from radiation damage to blood vessels, creates tissue that is weak and vulnerable to injury.^{15 16}

Risk factors

The most important risk factor for radiation-induced ORN is the dose of RT (increased risk demonstrated >60 Gy).¹⁵ Additionally, the location, stage and size of tumour are important; there is increase with larger and more advanced tumours. Bone that is closest to the tumour being irradiated is the most susceptible to ORN.¹ Surgeries such as osteotomy and mandibular resection can be used to debulk the tumour; these procedures, as well as procedures that occur after RT, including dental extraction due to poor oral hygiene, increase the risk of ORN.^{1 2}

Differential diagnosis

It is important to remember that ORN can present clinically similar to recurrent neoplasm.¹² Consequently, it is important to include ORN in the differential when working up a patient for suspicion of recurrent neoplasm in the setting of previous RT. The most common symptom among patients with hyoid ORN is odynophagia, which is also a common presenting symptom for recurrent head and neck cancer.^{3 7} Furthermore, odynophagia is also seen in mucositis, which can result from RT up to 2 months following treatment completion. Additional symptoms of patients with ORN include halitosis, and ear, jaw or throat pain; difficulties with chewing and swallowing can cause significant weight loss.^{3 7} More advanced disease may present with fractures, bone sequestration, trismus and fistulas, although haemoptysis has not been traditionally associated with ORN.¹⁷

Definitive diagnosis of ORN requires bone biopsy, which may commonly demonstrate bony necrosis and lack of tumour proliferation. Nevertheless, CT imaging can provide important clues to help differentiate ORN from malignancy. Alhilali¹⁸ evaluated 63 patients with ORN or tumour recurrence and reported that ORN was associated with an indistinct mass/inflammation, whereas tumour recurrence presented as a discrete cystic or solid mass. Bony sclerosis, when present, was only found in patients with ORN and never in patients with tumour recurrence. Moreover, positron emission tomography is shown to be unreliable for differentiating between ORN from recurrent tumour as there is overlap in the maximum and mean uptake between the two entities.

Treatment

Although historically hyperbaric oxygen therapy (HBOT) has been the mainstay of conservative treatment for ORN, its usefulness is currently considered controversial. In theory, the supplemental oxygen should stimulate bone growth and angiogenesis.³ However, much of the evidence for HBOT has been based on retrospective studies and literature reviews.¹⁵ Recently, a double-blinded randomised control trial demonstrated no therapeutic

Figure 2 Postoperative endoscopic view of the supraglottic region. The arrow points to the epiglottis for orientation. Note the absence of the hyoid bone. There is still pseudomembrane present, indicating ongoing tissue healing.

benefit of HBOT over placebo in recovery from mandibular ORN.¹⁹

The fibrotic-atrophy theory has facilitated the emergence of antioxidant treatments such as pentoxifylline and vitamin E as adjuvant therapeutic agents.²⁰ Phase II trials have demonstrated increased bone and mucosal healing with these interventions,²⁰ but randomised clinical trials are still required to establish efficacy of these treatments during the early stages of ORN.¹⁵ Nevertheless, recent treatment protocols have begun implementing these agents and have shown promising results. Multiple studies have shown that with conservative medical treatment (eg, pentoxifylline, vitamin E and antibiotics), progression past early stage ORN is rare.^{2 20 21} Furthermore, treatment initiated within a few days of bone exposure provides the greatest results.²

Recently, Lyons² proposed a four-stage system in which treatment with pentoxifylline, vitamin E and antibiotics cured stage 1 ORN (disease involving <2.5 cm of bone in asymptomatic patients) and halted progression of stage 2 ORN (disease involving >2.5 cm bone in asymptomatic patients). Individuals in stage 3 ORN (disease involving >2.5 cm of bone in symptomatic patients) required debridement, while stage 4 ORN (presence of fracture or fistula, or evidence of inferior dental nerve injury) warranted reconstruction with free flap.²

Although often necessary, debridement and bone resection can result in debilitating complications, including neurovascular injury and pharyngocutaneous fistula formation.³ To this end, endoscopic techniques have been entertained as a less invasive and safer surgical alternative. Indeed, endoscopic instruments are now widely available for resection of many oropharyngeal, hypopharyngeal, supraglottic, and glottic tumours; these instruments have also been used in the transoral resection of the hyoid bone. However, there are very few instruments currently available that can be placed down the shaft of a laryngoscope, while allowing for precise bone resection. Consequently, a growing number of fields have begun employing ultrasonic energy to precisely and safely complete endoscopic surgical procedures.⁴⁻⁷

Several studies have evaluated the value of the Sonopet Omni in the field of otolaryngology. In fact, a recent report has documented its value in performing laryngeal cartilage dissection.⁴ However, to the best of our knowledge, there is no report in the literature commenting on the use of the Sonopet for endoscopic removal of hyoid bone secondary to bone ORN.

The Sonopet Omni, originally developed in 1993, can emulsify, irrigate and aspirate bone⁷ by transmitting small ultrasound vibrations to its cutting tip, allowing for precise and controlled surgical incisions while minimising transmittance to contiguous structures.⁵ Studies have established the superiority of the Sonopet in manipulating bone and soft tissue when compared to conventional drills, demonstrating increased precision with decreased operative time and decreased blood loss.⁶ The instrument has been found to be efficacious in endoscopic skull base surgery, allowing the surgeon to carefully aspirate bone during trans-sphenoidal approaches and complex skull base tumour resections.⁶

In this particular case, we felt that the Sonopet would be useful for transoral resection of the hyoid due to the atraumatic nature of the instrument, and the proximity of the greater cornu of the hyoid to the superior thyroid and external carotid artery. Specifically, we felt the Sonopet would allow precise aspiration of the hyoid down to the base through the narrow corridor of an operating laryngoscope. The technique would minimise damage to the surrounding pharyngeal soft tissues, which

would not be possible with a standard extended cutting burr or other standard instruments.

Conclusion

This case demonstrates a patient with the unusual finding of hyoid ORN presenting with life-threatening haemoptysis in the setting of an oropharyngeal fistula. The fact that hyoid ORN can present clinically with symptoms similar to neoplastic recurrence has already been highlighted in several different articles. Once recurrence is ruled out, options for treatment include open and endoscopic techniques. We demonstrate the novel and effective use of the ultrasonic bone aspirator for resection of hyoid bone ORN through an endoscopic approach.

Learning points

- Osteoradionecrosis refers to a non-healing area of bone that has been present for at least 3 months; it may be associated with necrosis of overlying mucosa and/or skin.
- Osteoradionecrosis should be part of the differential when evaluating a patient with head and neck cancer for recurrent tumour after radiation therapy.
- Increasing evidence indicates that early stage osteoradionecrosis may be successfully cured or halted with aggressive and early medical management consisting of vitamin E, pentoxifylline and antibiotics.
- Ultrasonic aspirator technology can allow for successful, safer and less invasive surgical treatment of advanced osteoradionecrosis.

Competing interests None.

Patient consent Obtained.

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REFERENCES

- 1 Madrid C, Abarca M, Bouferrache K. Osteoradionecrosis: an update. *Oral Oncol* 2010;46:471–4.
- 2 Lyons A. Osteoradionecrosis: a review of current concepts in defining the extent of the disease and a new classification proposal. *Br J Oral Maxillofac Surg* 2014;52:392–5.
- 3 Silvestre-Rangil J, Silvestre FJ. Clinico-therapeutic management of osteoradionecrosis: a literature review and update. *Med Oral Patol Oral Cir Bucal* 2011;16:e900–4.
- 4 Daniero JJ, Brody RM, Fickes MG, et al. Ultrasonic surgical aspirator-assisted phonsurgery: a novel technique for laryngeal cartilage dissection. *Laryngoscope* 2014;24:1909–11.
- 5 Sivak-Callcott JA, Linberg JV, Patel S. Ultrasonic bone removal with the Sonopet Omni: a new instrument for orbital and lacrimal surgery. *Arch Ophthalmol* 2005;123:1595–7.
- 6 Baddour HM, Lupa MD, Patel ZM. Comparing use of the Sonopet(®) ultrasonic bone aspirator to traditional instrumentation during the endoscopic transsphenoidal approach in pituitary tumor resection. *Int Forum Allergy Rhinol* 2013;3:588–91.
- 7 Stryker Corporation. Stryker Sonopet ultrasonic aspirator. 1998–2013 (cited 2013 Oct 20) <http://www.stryker.com/en-us/products/Instruments/GeneralMultiSpecialtyInstruments/SonopetUltrasonicAspirator/139699>
- 8 Monceaux G, Périé S, Montravers F, et al. Osteoradionecrosis of the hyoid bone: a report of 3 cases. *Am J Otolaryngol* 1999;20:400–4.
- 9 Bhatia PL. Osteonecrosis of hyoid bone and thyroid cartilage. *Arch Otolaryngol* 1979;105:553–4.
- 10 Smith WK, Pfeleiderer AG, Millet B. Osteoradionecrosis of the hyoid presenting as a cause of intractable neck pain following radiotherapy and the role of magnetic resonance image scanning to aid diagnosis. *J Laryngol Otol* 2003;117:1003–5.
- 11 Robertson JS, Frauenhoffer EE, Stryker J, et al. Osteoradionecrosis of the hyoid induced by combined modality therapy for laryngeal carcinoma. *Ear Nose Throat J* 1995;74:578–81.

- 12 Yoo JS, Rosenthal DI, Mitchell K, *et al.* Osteoradionecrosis of the hyoid bone: imaging findings. *Am J Neuroradiol* 2010;4:761–6.
- 13 Meyer I. Infectious diseases of the jaws. *J Oral Surg* 1970;28:17–26.
- 14 Marx RE. A new concept in the treatment of osteoradionecrosis. *J Oral Maxillofac Surg (Internet)* 1983;41:351–7.
- 15 O'Dell K, Sinha U. Osteoradionecrosis. *Oral Maxillofac Surg Clin North Am (Internet)* 2011;23:455–64.
- 16 Delanian S, Lefaix JL. The radiation-induced fibroatrophic process: therapeutic perspective via the antioxidant pathway. *Radiother Oncol* 2004;73:119–31.
- 17 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.
- 18 Alhilali L. Osteoradionecrosis after radiation therapy for head and neck cancer: differentiation from recurrent disease with CT and PET/CT imaging. *Am J Neuroradiol* 2014;35:1405–11.
- 19 Annane D, Depondt J, Aubert P, *et al.* Hyperbaric oxygen therapy for radionecrosis of the jaw: a randomized, placebo-controlled, double-blind trial from the ORN96 study group. *J Clin Oncol (Internet)* 2004;22:4893–900.
- 20 Delanian S. Complete restoration of refractory mandibular osteoradionecrosis by prolonged treatment with a pentoxifylline-tocopherol-clodronate combination (PENTOCLO): a phase II trial. *Int J Radiat Oncol Biol Phys* 2011;80:832.
- 21 D'Souza J. Changing trends and the role of medical management on the outcome of patients treated for osteoradionecrosis of the mandible: experience from a regional head and neck unit. *Br J Oral Maxillofac Surg* 2014;52:356.

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