Osteoradionecrosis of the Mandible and Mastoiditis after Radiotherapy for Parotid Mucoepidermoid Carcinoma

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ABSTRACT:

Osteoradionecrosis of the mandible in conjunction with mastoiditis is an extremely rare occurrence following irradiation of salivary gland malignancy in the orofacial region. We report one such case of a patient who reported to us with trismus, jaw pain and ear discharge. Imaging of the jaws revealed classical features of osteoradionecrosis and mastoiditis. This case is important because, presenting features like trismus and dental infection lead us to investigative procedures that revealed extensive bone involvement including mastoiditis. Trismus progressively increased over a period of 8 years. In this case we would like to emphasise on the importance good oral hygiene in post radiotherapy stage for head and neck cancer.

KEYWORDS: Osteoradionecrosis; Mastoiditis; Radiotherapy; Parotid tumors
INTRODUCTION:

Osteoradionecrosis (ORN) is a condition of non-vital bone in a site of radiation injury which was first described by Marx in 1983 as hypovascularity, hypocellularity, and local tissue hypoxia.\(^1\). ORN can be spontaneous, but it most commonly results from tissue injury. The absence of reserve reparative capacity is a result of the prior radiation injury. Even apparently innocuous forms of trauma such as denture-related injury, ulcers, or tooth extraction can overwhelm the reparative capacity of the radiation-injured bone.

ORN is more common in the mandible but rare in the temporal bone.\(^2\) The first case of osteoradionecrosis involving the temporal bone was reported by Block in a patient with syringobulba in 1952.\(^3\) Co-occurrence of ORN in mandible as well as temporal bone have rarely been reported.

It is more common in the mandible than in the maxilla, probably because of the richer vascular supply to the maxilla and the fact that the mandible is more frequently irradiated.\(^4\)

We hereby report a rare instance of ORN involving the mandible and mastoid in a patient after radiotherapy for muco-epidermoid carcinoma of the parotid gland.

CASE REPORT:

A 38-year-old female patient reported to the Department of Oral Medicine and Radiology with the complaint of pain in the right back region of the lower jaw since 15 days. Pain was insidious in onset, continuous in nature and radiated to the right ear. Patient had restricted mouth opening. Patient also reported of intermittent discharge from the right ear since past 5 years, which was colourless initially and purulent later. Patient had visited
otolaryngologist for the ear discharge on numerous occasions, medications were prescribed but no reductions in symptoms were observed. Patient presented with history of surgery and radiotherapy in the same region for a nodular growth five years prior to the date of reporting. The discharge summary revealed the treatment modalities were carried out for a low-grade mucoepidermoid carcinoma. The patient reported of dryness of the mouth after surgery and radiotherapy. No histories of dizziness, tinnitus, vertigo or fainting episodes were reported.

On clinical examination bilateral temperomandibular joint movements were restricted with only 5 mm mouth opening. Right submandibular lymph node was palpable, enlarged, soft in consistency, tender and freely movable measuring about 3 cms in diameter (Figure 1).

Extra-oral examination revealed diffuse purplish black discoloration of the overlying skin in the pre-auricular, post-auricular region and angle of the mandible of the right side. The skin over the angle of the mandible had desquamated shiny appearance. A surgical scar extended from the right ear lobe to the base of the mandible. Purulent discharge was noticed in relation to the right external auditory meatus. There was diffuse areas of hyper and hypo pigmentation within the ear [Figure 2]. On palpation, the area was hard in consistency. The masseter muscle and the sternocleidomastoid muscle were fibrosed.

Intra-oral examination could not be performed due to restricted mouth opening. Orthopantamogram was made which revealed multiple decayed teeth in upper and lower arches. A mixed radiopaque-lucent area was observed distal to 45 extending upto the right condylar area. The upper and lower margins of the right body and the ramus appeared irregular and ill-defined. There was a discontinuity in the right lower border of the mandible near the angle region. A retained root stump was also observed in the same region surrounded by irregular patchy radiopaque and radiolucent areas. The path of the inferior alveolar canal
could not be traced through the lesion (Figure 3A). Right lateral oblique view of the body of the mandible revealed similar radiographical features (Figure 3B). Based upon the clinical, radiological features and the history of radiotherapy to the head and neck region, provisional diagnosis of osteoradionecrosis was made. Examination by an otolaryngologist revealed granulation of the tympanic membrane exposing the tympanic plate and necrosis of the overlying skin which was the cause of pus discharge from the ear.

Computed Tomography (CT) with contrast of the head and neck region was made which revealed right mandible, showing fluid within the marrow and multiple cortical breaks (figure 4). There was diffuse infiltrative oedema in the right masticator space. The right condylar area also showed marrow oedema with anterior erosion of the condyle which enhanced on post contrast scan (figure 5). The right mastoid cells showed fluid density consistent with inflammation (figure 6). There was no evidence of dural breach or extension of infection into the cerebellum or the middle cranial fossa.

Magnetic resonance imaging revealed findings consistent with CT (Figure 7, 8, 9). The right side of the tongue showed fatty atrophy but the posterior aspect showed high signal intensity on T2 and STIR images, there is likely extension of the inflammatory process from the mandible. There is hypertrophy of the left half of the tongue and the asymmetry being secondary to post radiotherapy appearances. Right submental, bilateral submandibular, left level II, III and level V lymph nodes were seen. In view of the post radiotherapy for right parotid malignancy, the results were suggestive of osteoradionecrosis with soft tissue infection and right mastoiditis with no evidence of extension of infection into the brain.

Extraction of residual root stump which was the probable source of infection for the ORN was carried out under local anaesthesia. Patient was administered empirical antibiotic
therapy. Since we did not have the expertise to further manage the complicated case we referred it to higher centres for treatment.

Initially Culture and Sensitivity tests were carried out for the pus discharge from the right ear. Following this a course of IV Pencillins were administered along with antibiotic ear drops. Patient was further referred to higher centre for Hyperbaric oxygen therapy. Patient at present is asymptomatic.

DISCUSSION:

Chronic otomastoiditis may result as a complication arising from osteoradionecrosis of the temporal bone as in our case. This may further result in hearing loss, gross tissue extrusion, more severe complications including meningitis, facial palsy, intradural and/or extradural abscesses, pneumocephalus, lateral sinus thrombosis, fistula formation into the parotid gland or temporal mandibular joint, and other cranial neuropathies. In our case the right temporomandibular joint was involved.

In our patient, the disease was relatively diffuse involving mostly the mastoid, the mandible including the condyle and masticator space, but not the cranial base. The patient presented with an exceptionally rare complication of osteoradionecrosis of the temporal bone, secondary to radiotherapy for parotid tumor. On consultation with a otolaryngologist, tympanic membrane perforation noticed. However, considering the lack of otologic complaints before radiotherapy, the perforation was thought to have been caused by radionecrosis and/or subsequent infection.
The incidence of temporal bone ORN is higher after mastoidectomy for facial nerve identification or resection in patients undergoing parotidectomy with postoperative radiotherapy.\textsuperscript{6}

The management of osteoradionecrosis in the temporal bone is controversial. In the localized type, conservative treatment with frequent aural cleansing and topical antibiotics is often administered.\textsuperscript{7}

Oversew of the ear canal with mastoid obliteration should be considered in this subgroup of patients to avoid this long-term complication of radiotherapy used in the treatment of malignant parotid tumors.\textsuperscript{6}

The risk for osteoradionecrosis and infection can be minimized by removing all poorly supported teeth, allowing sufficient time for the extraction wounds to heal before beginning radiation therapy, and adjusting dentures to minimize the risk for denture sores.\textsuperscript{4}

Prior to beginning radiation therapy, all patients should undergo a thorough dental evaluation, including full mouth radiographs, dental and periodontal diagnosis, and prognosis for each tooth. It is generally accepted that meticulous preventive dental treatment should be planned for patients receiving radiotherapy to the head and neck region.\textsuperscript{8} In addition to oral hygiene maintenance with routine dental follow-up, non-surgical or ‘conservative’ therapy includes nutritional support, topical medicaments, systemic antibiotics and hyperbaric oxygen.\textsuperscript{8} In 1983, Marx proposed a staging protocol (which was modified subsequently) combining surgery and hyperbaric oxygen for more aggressive treatment of ORN. He was able to achieve complete resolution in all 58 cases.\textsuperscript{9}

The management of patients afflicted with ORN of the jaws usually consists of a combination of different modalities and will be determined by factors such as the size of the
defect, the signs and symptoms of the patient, the cosmetic and functional derangement consequent to the complication.\textsuperscript{8} For an unresponding case or the diffuse type of osteoradionecrosis, surgical management is indicated. Surgical management of osteoradionecrosis has met with limited success because of the difficulty of accurate assessment of the viability of non-necrotic bone. Failure to resect all non-viable bone results in recurrence of a necrotic focus.\textsuperscript{10} Since such expertise was not available in our case, the patient had to be referred to higher centre for further management.

Since the introduction of Marx's protocol, there have been advances in surgical techniques (i.e. microvascular surgery), as well as in imaging techniques, which have significantly impacted on the diagnosis and management of ORN. High resolution CT scans and orthopantamograms have become a key component in evaluating and staging ORN, prior to formulating a treatment plan. Reconstructions are now routinely performed immediately after resection of the diseased tissue rather than in a staged fashion.\textsuperscript{11} For advanced ORN of the mandible, radical resection followed by reconstruction using free flap provides a reliable means of obtaining good wound healing with acceptable aesthetic and functional results.\textsuperscript{12}

Despite diligent radical treatment, suspicion of recurrent cancer always persists. Extensive osteoradionecrosis with a multiple discharging fistula, a large area of exposed necrotic bone, or a coexistent fracture should be treated primarily with radical sequestrectomy and microvascular free flap reconstruction. Surgery plays a major role in controlling osteoradionecrosis along with adjuvant hyperbaric oxygen therapy.\textsuperscript{13}

With the advent of highly precise conformal therapies, such as IMRT, the accurate localization and delivery of radiotherapy is increasingly important. Recent advances in image-guided radiotherapy provide increased tumor localization by improving the identification of areas of tumor burden, by minimizing the effects of patient setup errors
caused by intra-/interfraction motion, and by allowing for adaptive replanning to changes that occur in the tumor or patient during long courses of radiotherapy. In doing so, these changes are leading to improvements in the therapeutic ratio, where doses are increased at diseased-sites and minimized at normal tissues.\textsuperscript{14}

We must also keep in mind that no single imaging modality can provide an entire picture of tumour profiling. Anatomical and functional imaging fusion is essential. Future studies should help determine whether incorporating these and other imaging modalities will improve our ability to cure head and neck cancer and/or reduce the morbidity of the treatment.\textsuperscript{15}

**CONCLUSION:**

Data available regarding osteoradionecrosis are insufficient to assist clinicians faced with the management and in counselling patients as to the anticipated outcome of therapy. Efforts should be made to monitor patients who have been enrolled in such treatment protocols for long periods of time to evaluate the quality of life and late complications of therapy.

This report stresses the importance of educating health care professionals, especially dentists and oral surgeons, of a rare but important side effect of radiation therapy in the head and neck area. It is especially useful in bringing to light the importance of dental evaluation prior to and after radiation therapy to improve patient treatment outcome.
REFERENCES:


FIGURE LEGENDS

Figure 1: Enlarged right submandibular lymphnode.

Figure 2: Diffuse purplish black discoloration of the overlying skin and pus discharge from the right external auditory meatus.
Figure 3A: Orthopantamogram showing patchy radioopaque-radiolucent areas in the area of right body and ramus of the mandible

Figure 3B: Right lateral oblique showing extension of lesion into the ramus
Figure 4: Axial CT section showing right mandibular cortical breach

Figure 5: Axial CT section showing anterior erosion of the right condyle
Figure 6: Axial CT section showing fluid density of right mastoid

Figure 7: Axial MR section showing fluid within marrow, multiple cortical breaks
Figure 8: Axial MR section showing marrow edema around right condylar head

Figure 9: Axial MR section showing fluid density within the right mastoid consistent with inflammation.