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Radiology in Focus

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

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Abstract
Osteoradionecrosis of the hyoid has been reported rarely in the worldwide literature. We present the case of a 56-year-old gentleman who presented with intractable neck pain, following surgery and radiotherapy for a $T_2N_{2c}M_0$ tongue base carcinoma, to highlight the need to consider osteoradionecrosis of the hyoid rather than recurrence of the carcinoma as the cause of such symptoms. The previously unreported appearance of osteoradionecrosis of the hyoid on a magnetic resonance image (MRI) scan and the use of this investigation to aid diagnosis is discussed.

Key words: Hyoid Bone; Osteoradionecrosis; Magnetic Resonance Imaging

Introduction

Osteoradionecrosis is the *in situ* death of a segment of bone following radiotherapy. Histological examination reveals osteocytonuclei, osteoclastic resportion of bone and fibrosis of the marrow space.[1] The pathological changes of osteoradionecrosis are due to both hypoxia related to the microvascular changes as well as damage to the osteoblasts as a result of the irradiation. This prevents bony repair after any further trauma resulting in osteoradionecrosis.

Combined treatment is used in the management of head and neck malignancies and each method, radiation, chemotherapy and surgery, may contribute to the development of osteoradionecrosis. Surgery via local tissue trauma may reduce the vascularity of the hyoid. Radiation however, especially when increasing dosages are employed, is thought to be the essential factor.[2] This accounts for the risk of developing osteoradionecrosis being higher when the tumour lies closer to the bone. The hypoxic hypocellular tissue results in tissue breakdown and a chronic absence of wound healing.[3] Other risk factors include the T stage, type of irradiation, heavy alcohol consumption and smoking as well as the patient's nutritional status.[1]

MRI scans are increasingly employed in the assessment of the soft tissues in head and neck malignancies and the MRI appearance in osteoradionecrosis of the mandible, temporal bones and pelvis has been documented. Osteoradionecrosis of the mandible and temporal bone has been extensively reported in the literature but osteoradionecrosis of the hyoid appears to be much rarer. Four papers reporting six cases document this condition.[1,4–6] We report a further case affecting the hyoid where the differential between osteoradionecrosis and recurrent disease presented a diagnostic dilemma that was clarified by MRI scan. This has not previously been reported and its value is discussed.

Case report

A 56-year-old man presented with asymptomatic bilateral upper deep cervical lymphadenopathy which had been present for some months. He had no associated ear, nose or throat complaints. He had smoked 20 cigarettes a day for most of his life.

Examination revealed bilateral jugulodigastric lymphadenopathy (more marked on the left) but no obvious ENT primary. Fine needle aspiration biopsy did not reveal any malignant cells therefore a left cervical lymph node biopsy was carried out revealing a metastatic squamous cell carcinoma. On subsequent pharyngolaryngoscopy a small area of friable tissue in the right tongue base was identified as the primary site and a modified left neck dissection of levels I to IV with preservation of the accessory nerve was also carried out. Histology confirmed other involved nodes with evidence of extracapsular spread.

A subsequent MRI scan demonstrated a 2×3 cm tumour in the right tongue base as well as multiple enlarged lymph nodes in the right upper deep cervical region and therefore the patient had radical radiotherapy to the primary site and both sides of the neck. This was given in two phases; phase 1 involved 17 Gy in two fractions over three days using 6 mV photons and phase 2 was 36 Gy in 18 fractions over four weeks using 6 mV photons. He developed a marked radiation reaction.

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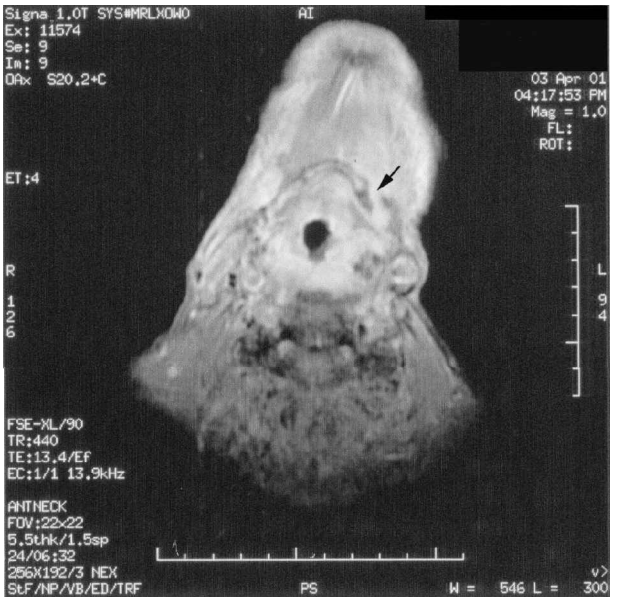

FIG. 1

MR T2WI (after gadolinium administration) of patient showing hyperintensity of the osteonecrotic hyoid bone.

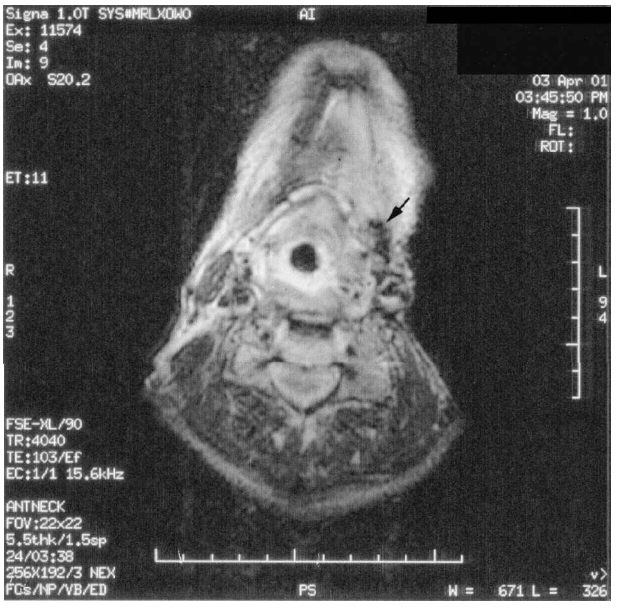

FIG. 2

MR T1WI of patient (after gadolinium administration) showing hypointensity of the osteonecrotic hyoid bone.

- **This is a report of osteoradionecrosis of the hyoid bone following radiotherapy for tongue base carcinoma**
- **The differential diagnosis was between recurrent tumour and osteoradionecrosis**
- **Osteoradionecrosis of the hyoid has, rarely, been reported previously**
- **This case is added as magnetic resonance imaging proved helpful in distinguishing the aetiology of the problem. The value of this imaging modality is highlighted**

Eight months following radiotherapy, he began to complain of severe pain in the left side of his throat radiating to his ear and despite a combination of MST continuous® and codeine phosphate he experienced marked swallowing difficulties resulting in a significant weight loss that was throught to be due to tumour recurrence. A MRI scan showed high signal on T2WI (Figure 1) and absence of enhancement on T1WI (Figure 2) sequences after gadolinium administration within both the soft tissues and hyoid bone. This would be in keeping with osteoradionecrosis with inflammation (clinically apparent at the time of the MR study), rather than neoplasm. A necrotic ulcer and exposed hyoid was found at the apex of the left piriform fossa but repeated biopsies only revealed changes compatible with previous irradiation but no evidence of malignancy. In view of this and the changes on MRI a diagnosis of osteoradionecrosis of the hyoid as the cause of his symptoms was made.

His symptoms resolved following local endoscopic removal of the involved hyoid bone. The bone and surrounding soft tissue was sent for histological examination which showed radiation changes only with no evidence of malignancy. Unfortunately the patient died suddenly at home. The exact cause of death was not established since no autopsy had been requested.

Discussion

Osteoradionecrosis presents a diagnostic dilemma since recurrent or persistent neoplastic disease may present in similar fashion and must be ruled out. Although patients with an aseptic osteoradionecrosis may remain asymptomatic,[7] most patients present, like ours, with pain together with signs of inflammation. The most common symptom when the hyoid is affected is odynophagia resulting in a patient in poor physical condition with significant weight loss. Others had dysphagia, ear pain, pharyngeal discomfort, pooling of saliva, and a lateral cervical mass +/− fistula. In all reported cases, shallow ulcers with sequestered parts of the hyoid bone visible in the pharyngeal lumen were found which in this case offered a convenient means of removing the diseased bone.

The dose of radiotherapy given to this patient was unusual as he received two very high dose fractions (of 17 Gy in two fractions over three days with 6 MV photons) initially, although this was a standard regimen for the protocol used at that time. The use of such high doses of radiotherapy is a definitive risk factor for osteoradionecrosis which is an observation that has previously been discussed in the literature.[2]

Repeated biopsies showed no evidence of neoplasm. We feel that it is essential to biopsy when there is a high degree of clinical suspicion that tumour is present, although repeated biopsies may in fact precipitate osteoradionecrosis by providing the traumatic catalyst.[8] However osteoradionecrosis and tumour may co-exist and indeed the presence of tumour may expose irradiated bones and cause osteoradionecrosis.[4]

201 thallium, 99m technetium, positron emission tomography (PET), computed tomography (CT), MRI, scans have all been employed to try and determine whether patients presenting with the symptoms discussed have persistent or recurrent tumour or whether osteoradionecrosis has occurred. Monceaux reported that using scintigraphic examination with ^{201}thallium revealed that uptake suggests the presence of residual disease whilst bone marker ^{99m}technetium-diphosphonate uptake would be indicative of osteonecrosis.[1]

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PET is a relatively new imaging technique using ^{18}FDG, a fluorine-labelled glucose analogue, and its use in the detection of tumour persistence or recurrence post-treatment of head and neck malignancies is currently being evaluated. An 86 per cent sensitivity and 93 per cent specificity with an overall accuracy of 89 per cent in distinguishing tumour from radiation-induced changes in the neck was reported by Farber *et al.*[9] Li *et al.*[10] reported an overall accuracy of 88 per cent with FDG PET compared to 66 per cent for CT and/or MRI. From these and other studies[11] it appears that PET perhaps with PET-MRI coregistration techniques will be useful in the detection of tumour.

Some studies have shown that the CT findings are not specific and have not been able to differentiate reliably between recurrent tumour and osteoradionecrosis.[6] A review of the literature describing the MR appearance of osteoradionecrosis in bones other than in the hyoid agree that osteoradionecrosis results in a hypointensity on T1WI but the appearance of T2WI appears to vary. Chong *et al.*[12] found that their MRI scans showed T2 hyperintensity of involved osteoradionecrotic bone whereas Store *et al.*[13] did not. This apparent discrepancy in the T2WI is thought to be due to the presence or absence of inflammation associated with the osteoradionecrosis at the time of the MR scan.[14] Since inflammation commonly accompanies osteoradionecrosis MR scanning is still useful in the differentiation from tumour which unlike osteoradionecrosis shows enhancement on T1WI and has the additional advantage over CT, thallium and technetium scans of not exposing the patient to radiation.

It is important to identify those patients with osteradionecrosis rather than tumour since the treatment of this condition is different. Surgery can be less radical in the form of debridement. The use of intravenous antibiotics and hyperbaric oxygen have been advocated by Robertson[6] although Bhatia[4] treated his patient with cyclophosphamide.

Conclusion

The usefulness of MR scanning and importance of repeated biopsies in differentiating between osteoradionecrosis and neoplasm has been highlighted in this patient. The use of PET-MRI coregistration techniques may improve diagnosis, which is essential if appropriate treatment is to be given and radical surgery is to be avoided.

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