Radiation-induced osteosarcoma of the maxilla

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ABSTRACT

Radiation-induced sarcomas are well-known though uncommon potential late sequelae of radiation therapy. We report irradiation-induced osteosarcoma involving the maxilla following treatment of nasopharyngeal carcinoma in a 44-year-old man who presented with painful cheek swelling. Radiographs and computed tomography showed a large destructive lesion of the left maxilla. Diagnosis of osteosarcoma was confirmed by excision biopsy.

Keywords: computed tomography, maxillary osteosarcoma, nasopharyngeal carcinoma, osteosarcoma, radiation-induced sarcoma

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INTRODUCTION

Radiation-induced osteosarcoma arises as a late complication of radiation treatment. These tumours are uncommon but are aggressive, occurring in bones in an irradiated area after a latent period of five or more years following radiotherapy. Histological proof of the tumour is necessary to distinguish it from other radiotherapy changes such as osteonecrosis. Long-term patient follow-up and a high index of suspicion are crucial for early detection and timely intervention. In this report, we present a 44-year-old man with osteosarcoma of the maxilla diagnosed 14 years after radiotherapy treatment of nasopharyngeal carcinoma.

CASE REPORT

A 44-year-old man presented to our hospital for pain and swelling over the cheek that was increasing in size and advancing towards the eye. Examination showed an expansile mass in the left maxillary region with proptosis of the left eye. There was paraesthesia of the left side cheek, left nasal obstruction, and blurring of vision in left eye. The overlying skin appeared normal (Fig. 1). Radiographs showed a destructive bony lesion of the maxilla with exuberant new bone formation and spiculated margin (Fig. 2). Computed tomography (CT) demonstrated a large bony mass in the left maxilla completely destroying the left antrum. The bony mass extended into the left orbit, left temporal bone, as well as the nasal cavity (Fig. 3). A preliminary diagnosis of osteosarcoma

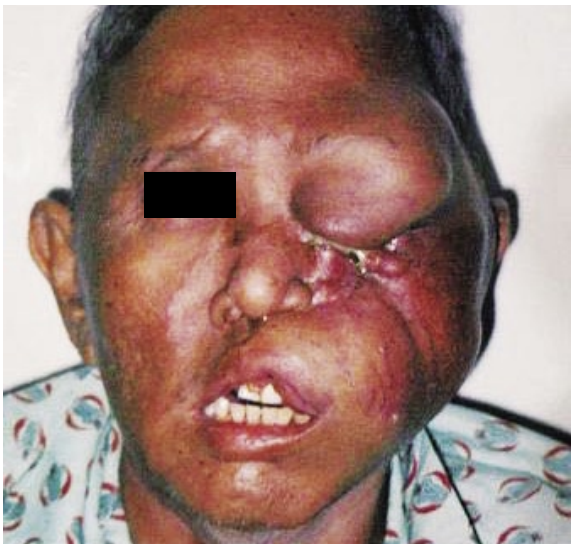

Fig. 1 Clinical photograph shows a painful expansile mass in the left maxillary region with proptosis of the left eye.

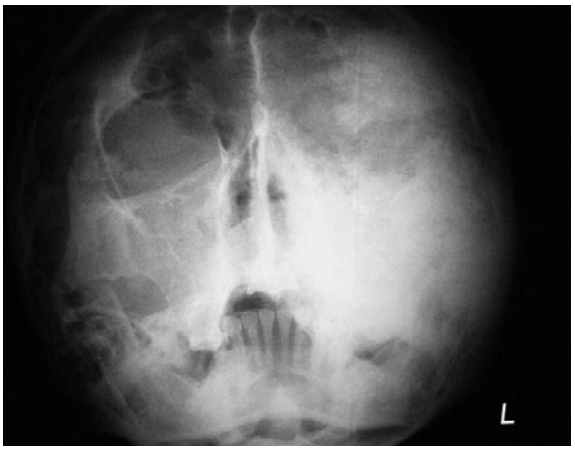

Fig. 2 AP radiograph shows a large mixed lytic and sclerotic lesion in the left maxilla with a spiculated margin.

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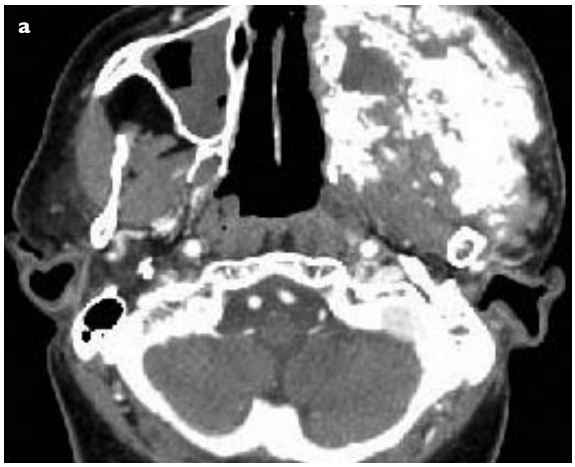

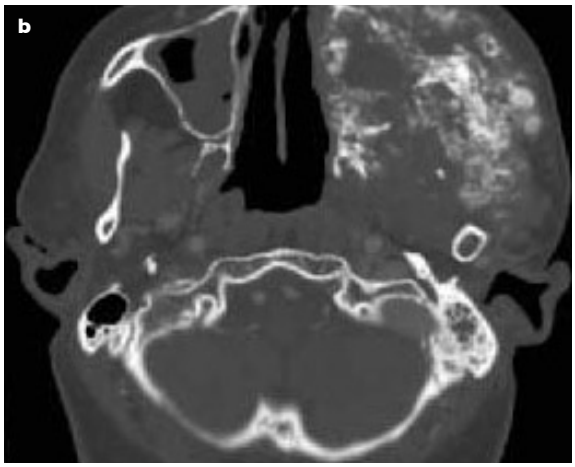

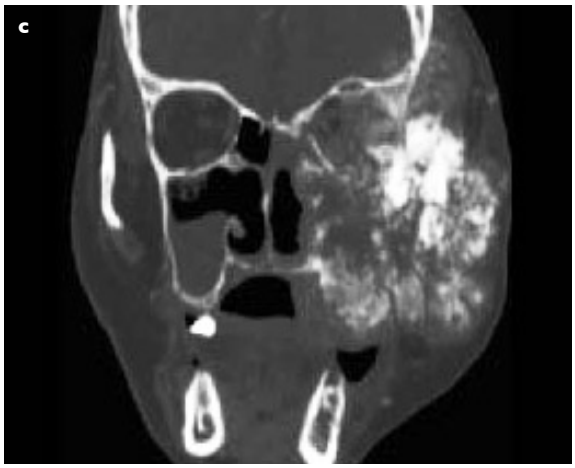

Fig. 3 Axial (a) soft tissue and (b) bone window, and (c) coronal bone window CT images show a large bony mass in the left maxilla that completely destroys the left antrum. The bony mass extends into the left orbit, left temporal bone, as well as the nasal cavity.

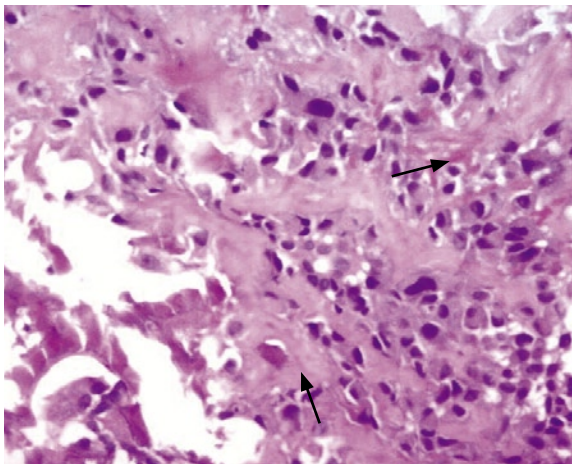

Fig. 4 Photomicrograph of the biopsy specimen shows a neoplasm composed of sheets of spindle cells having eosinophilic cytoplasm and hyperchromatic nuclei. Layering of osteoid by the tumour cells is seen (arrows). Scattered mitotic figures are noted (Haemotoxylin & eosin, ×200).

of the maxilla was made which was confirmed by excision biopsy (Fig. 4). CT of the lung revealed no lung metastases.

On further questioning, patient was diagnosed to have nasopharyngeal carcinoma (NPC) 14 years ago. He was previously treated with radiation in Indonesia though more specific details of his treatment were not forthcoming. He was well since, until he noticed a swelling of the left cheek one year ago but had opted for traditional treatment. Because there was no improvement, the patient opted to come to Malaysia for further treatment. He was given three cycles of chemotherapy. The CT appearances after chemotherapy were unchanged from the admission scan. Unfortunately, he had developed electrolyte imbalance due to complications of chemotherapy. He had then returned to Indonesia and was lost to follow-up.

DISCUSSION

Nasopharyngeal carcinoma (NPC) is one of the most common types of cancer in Southeast Asia. Radical radiotherapy with a dosage of 65 to 70 Gy for six to seven weeks is the main treatment modality for the primary tumour. If there is cervical lymph node involvement, the neck receives an additional 50 to 60 Gy subsequently over the next five to six weeks[1]. When radiotherapy is used with a curative intention, the possibility of the appearance of late complications in the irradiated tissues must be considered. The spectrum of malignant neoplasms secondary to radiation is wide, and includes tumours of the skin, thyroid and bone[2]. Because radiation fields for NPC may include the maxillary bone, mandible bone, pterygoid bone, and the skull base, radiation-induced osteosarcoma can arise from these sites as one of the late complications of radiotherapy.

The radiation fields for nasopharynx normally include two opposite preauricular and prenasal fields. For patients with nasal cavity or poststyloid area invasion, an anterior or post-auricular supplementary field is added. Most sarcomas have been reported to occur after exposure to 55 Gy, with the dose ranging from 16 to 112 Gy[1]. The latent time of

radiation-induced osteosarcoma after irradiation for NPC ranges from four to 27 years, with a mean of 13.3 years[3]. There have only been eight articles concerning radiation-induced osteosarcoma in NPC reported in the English literature, and all but one are case reports[1].

The appearance of sarcomas in irradiated tissues is an uncommon, but well-documented, long-term complication of radiotherapy treatment. The incidence of radiation-induced tumours is increasing in the oncology patient population, as a result of increased overall survival, because of the improvement in the efficacy of cancer treatment[1,3,4]. The largest study reported in the English literature by Liu et al described 15 patients with NPC, and found the location where the radiation-induced osteosarcoma arose as one of the late complications of NPC, which included: 33.3% from the maxilla, 46.7% from the mandible, and 20% from a mixture of the nasal cavity and paranasal sinuses[1]. They estimated the incidence rate of radiation-induced osteosarcoma in NPC to be approximately 0.037%[1].

Although the pathogenesis is unknown, various predisposing factors have been proposed. It is suggested that the patients who harbour the mutation in tumour suppressor genes like p53 and retinoblastoma gene, are more prone to develop radiation-induced osteosarcoma. Furthermore, children appear to be more susceptible than adults[4]. Post-radiation osteogenic sarcomas of the facial bones have been reported in patients who had benign diseases such as fibrous dysplasia or Paget's disease, and had been treated with radiotherapy in the past[2]. A soft tissue mass, destruction of bone, and tumour due to new bone formation are the main characteristics on radiography and CT. The degree of new bone formation on CT varies from none to a large amount.

The most common clinical finding of the head and neck radiation-induced osteosarcoma is pain and swelling due to the bone tumour, and there may be a pathological fracture. The initial treatment is radical surgical removal if feasible, because it offers the only possibility of curative treatment, as chemotherapy and radiation are associated with poor response. However, it has been emphasised that early detection of radiation-induced tumours is important though difficult, as changes seen after radiotherapy have been usually attributed to other causes such as osteonecrosis[2]. In 1948, Cahan et al, in describing 11 cases of sarcomas arising from irradiated bones, established four criteria for the diagnosis of radiation-induced osteosarcoma that are still valid today. These are: histological or radiological proof that there was no previous tumour in the involved bone, development of sarcoma in an irradiated area, latency period of at least five years or more and histological confirmation of the diagnosis[5]. The patient in our case report fulfils the above-mentioned criteria.

The prognosis of radiation-induced sarcoma is generally thought to be worse than primary sarcomas, regardless of site. Most series report overall five-year survival rates in the range of 10% to 30%[1]. Several factors account for this poor survival rate. Often, there is a delay in diagnosis until an advanced stage of disease because of the difficult distinction between neoplastic and radiation changes. To prevent radiation-induced sarcoma, it is important to be meticulous to give the appropriate radiation dose in carefully-planned radiation fields. Because of the aggressive nature of radiation-induced sarcoma, careful long-term follow-up of radiated patients is crucial. A high index of suspicion is needed for early detection and timely intervention, which could result in a less disfiguring surgery.

In conclusion, radiation-induced sarcomas are potential late sequelae of radiation therapy. These tumours are very aggressive and often elude early detection, thereby hindering timely intervention. Although the condition is rare, the possibility of radiation-induced sarcoma must be considered in any patient who had received radiation treatment.

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