Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology 26 (2014) 590–595

Contents lists available at ScienceDirect

Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology

journal homepage: www.elsevier.com/locate/jomsmp

Case Report

Low grade osteosarcoma of maxilla: Report of a case and review of literature

Biyas Bhowmik[a,*], Venkatesh G. Naikmasur[a], Swetha Acharya[b], Amsavardani Tayaar S[b], Rashmi Bangera[b], U.S. Dinesh[c]

[a] Department of Oral Medicine and Radiology, S.D.M. College of Dental Sciences and Hospital, Dharwad, Karnataka, India
[b] Department of Oral Pathology, S.D.M. College of Dental Sciences and Hospital, Dharwad, Karnataka, India
[c] Department of General Pathology, S.D.M. College of Medical Sciences and Hospital, Dharwad, Karnataka, India

ARTICLE INFO

Article history:
Received 5 February 2013
Received in revised form 21 August 2013
Accepted 18 September 2013
Available online 30 October 2013

Keywords:
Computed Tomography (CT)
Immunohistochemistry
Low-grade osteosarcoma (LGOS)
Painless swelling of maxilla
Paresthesia
Spindle cell lesions

ABSTRACT

Osteosarcoma is a rare malignant bone tumour characterized by bone or osteoid formation within. Low-grade osteosarcoma (LGOS) represents less than 1% of all osteosarcomas in the body. Because of its rarity and well differentiation, LGOS is usually misdiagnosed as a benign lesion. A 21-year-old male patient presented with a history of slowly expanding hard, painless mass in the left maxilla since six months, along with difficulty in breathing and numbness over left side of the nose since last two months. The radiological and histologic findings summatively suggested a spindle cell tumour with a wide range of differential diagnoses. This manuscript presents a critical evaluation of the clinical, radiographic, histologic and immunohistochemical findings of the case with a detailed discussion of all the probable differential diagnoses.

1. Introduction

Spindle cell lesions of head and neck are diverse with clinical and biological heterogeneity [1]. What makes such cases even more diagnostically challenging is the fact that for biopsies from the head and neck, the range of tissues is varied but the amount of material made available to the histopathologist is limited [2]. Many are aggressive in nature making histopathologic discrimination between benign and malignant lesions important. The spindle cell lesion further poses a problem, when calcifications are evident [1]. Osteogenic sarcoma/osteosarcoma (OS) is one such entity that comprises of spindle cell component and bone deposition.

OS of the jaw is a very rare tumour of the head and neck comprising of 1% of all malignant tumours of head and neck [3]. Strictly from the histologic point of view, OS comprises a family of lesions with considerable diversity in histologic features and grade. It is a malignant tumour of mesodermal or connective tissue origin within which the tumour cells produce bone or osteoid. Although it may appear as though the cells giving rise to OS must be of osteoblastic derivation, there is no evidence that osteoblasts after differentiating from osteoprogenitor cells, can actually revert to more primitive cells, let alone malignant ones. This implication is inherent in the more arcane term osteogenic sarcoma, which however has fallen into disuse [4]. Due to their complexity in clinical features, radiography and the rarity in daily clinical practice, the preoperative diagnosis of OS is often difficult.

The purpose of this case report is to highlight the importance of diagnosing a malignant lesion that mimics a benign lesion histologically with the aid of imaging techniques to arrive at the right diagnosis. This presentation focuses on how even selecting the most appropriate set of investigations can sometimes not help us in pinning down the diagnosis.

2. Case report

A 21-year-old male patient reported with a complaint of hard, painless swelling in the left maxilla since six months and difficulty in breathing and numbness over left side of the nose since two months. The swelling had progressively increased in size and previous medication did not improve his condition. The family dentist had advised an Orthopantomograph one month ago, following which two teeth were extracted. One of it was supposed to be decayed and the other mobile. Nevertheless, the swelling did

☆ AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

* Corresponding author. Tel.: +91 08348067713.
E-mail addresses: biyas9bhowmik@gmail.com (B. Bhowmik), drvnmasur@gmail.com (V.G. Naikmasur), sbacharya@gmail.com (S. Acharya), amsavardani@yahoo.co.in (A. Tayaar S), cutestar3012@yahoo.com (R. Bangera).

2212-5558/$ – see front matter
http://dx.doi.org/10.1016/j.ajoms.2013.09.006

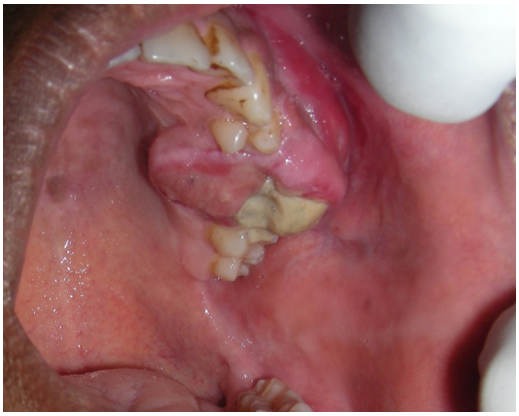

Fig. 1. Intraoral view of low-grade osteosarcoma of maxilla showing a polypoid growth in the left maxillary premolar-molar region with buccal cortical plate expansion, missing 25 and 26, and sequestrum formation at the site of extraction.

not regress. Extra-oral examination revealed a diffuse, hard and non-tender swelling on the left middle third of the face. A left submandibular lymph node was palpable and tender. Intra-orally, a non-tender, firm to hard, polypoid growth was evident, measuring 4 cm × 5 cm in the left maxillary premolar-molar region with buccal cortical plate expansion. 25 and 26 were clinically missing and there was sequestrum formation at the site of extraction and 24 was grade I mobile (Fig. 1). The lesion was provisionally diagnosed as an aggressive odontogenic tumour. Necessary radiographs and laboratory investigations were advised.

Orthopantomograph revealed a well-defined relatively radiopaque shadow in the left maxilla extending from 24 to 27, suggestive of soft tissue mass. 25 and 26 were missing (Fig. 2A). Maxillary occlusal radiograph showed irregular specks of calcification and buccal cortical plate expansion (Fig. 2B). Paranasal sinus view showed an irregular ill-defined radiopacity infiltrating the left maxillary sinus. The lesion was assumed to be an aggressive/atypical fibro-osseous reaction. The clinical features like numbness and difficulty in breathing remained less understood at this stage along with the extent of the lesion and its relation to maxillary sinus. To supplement this, a Computed Tomography (CT) scan was advised. CT revealed a fairly well-defined heterogeneous osteolytic lesion in maxillary alveolus with cortical thinning and erosion of bone from 22 to 27 (Fig. 2C and D). Superiorly, it extended into the maxillary sinus through focal erosions in the floor of the sinus, laterally up to the zygomatico-maxillary buttress whereas inferiorly, it extended into the palatine process of the maxilla and the hard palate. Numerous specks of calcifications within the lesion, superior displacement of inferior turbinate and deviation of nasal septum to the right were evident. Impression was that of an aggressive odontogenic tumour.

Further a biopsy was mandated to identify the lesion. An incisional biopsy revealed only reactive hyperplastic tissue with inflammation and few spindle cells following which the procedure was advised to be repeated. A possibility of inflammatory myofibroblastic tumour was considered. Surgical excision of the tumour with wide surgical margins was performed. Post-operatively the patient was put under a course of antibiotics for uneventful recovery.

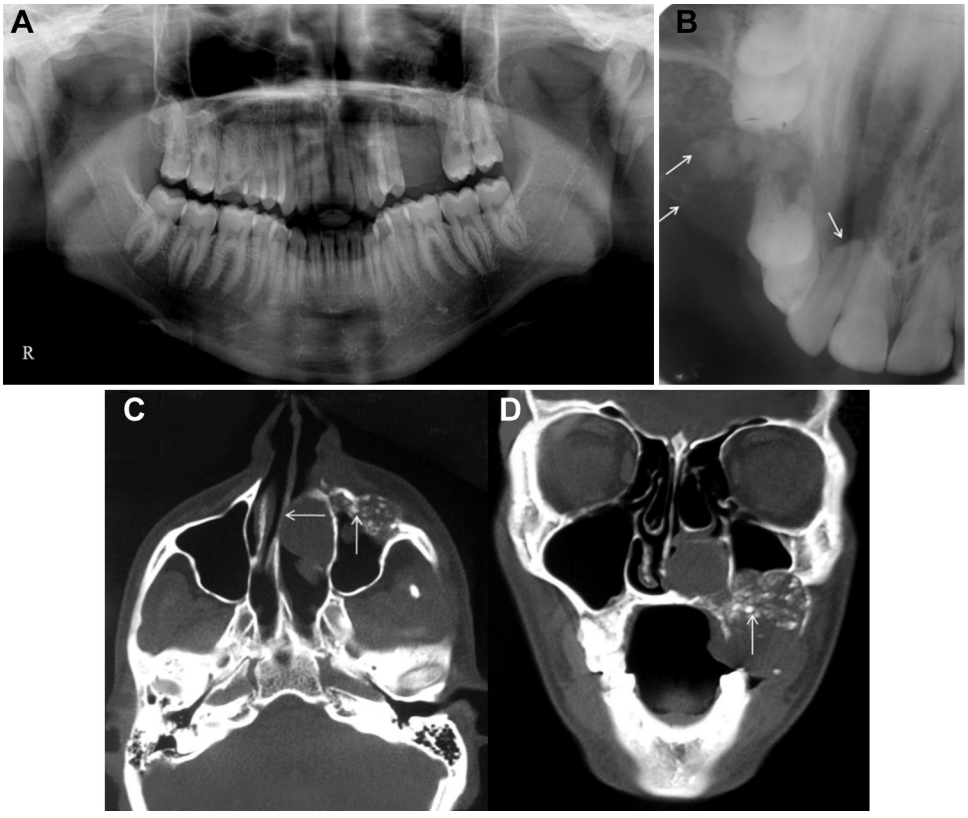

Fig. 2. Radiographic findings of low-grade osteosarcoma of maxilla. (A) Orthopantomograph showing a well-defined relatively radiopaque shadow in the left maxilla extending from 24 to 27, suggestive of soft tissue mass, and missing 25 and 26. (B) Maxillary occlusal radiograph showing irregular specks of calcification and buccal cortical plate expansion. Periapical resorption of 22 seen. (C, D) CT axial and coronal sections revealed a fairly well-defined heterogeneous osteolytic lesion in maxillary alveolus with cortical thinning and erosion of bone from 22 to 27. Superiorly, it extended into the maxillary sinus, laterally up to the zygomatico-maxillary buttress, and inferiorly into the palatine process of maxilla and the hard palate. Numerous specks of calcification within the lesion, superior displacement of inferior turbinate and deviation of nasal septum to the right were also evident.