Case Report

Osteosarcoma of the Mandible: A Case Report and Short Literature Review

Abstract

Osteosarcomas comprise of a heterogeneous group of tumors with a predilection for involving long bones commonly. The jaw remains a rare site of involvement, and these tumors can be considered a distinct entity. The key for managing them lies in histological and radiological correlation for an early diagnosis. Surgery and chemotherapy remain the main modalities of treatment. We present a case of osteogenic sarcoma of the mandible that after radical treatment had local recurrence and distant metastasis.

Keywords: *Mandible, metastasis, osteosarcoma*

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Introduction

Osteosarcoma (OS) is the most common primary malignant bone tumor in children and adults[1] and accounts for about 20% of all sarcomas.[2] It commonly affects the appendicular skeleton, whereas the maxillofacial region is a rare site seen in only around 6.5% cases.[3] The median age of such patients is between 34 and 36 years, which is greater than the median age of patients of OS of the long bones by about 15 years. These tumors have a slight predilection toward the male sex, and they are sometimes also referred to as gnathic OSs.[4-6] There have been around 300 cases of OS of head-and-neck region from all over the world reported in PubMed indexed journals spanning from 1967 to 2010.[7]

We present a rare case of OS of the mandible which was treated radically with neoadjuvant chemotherapy and surgery. We also follow the clinical course of the disease over a period of 5 years. The uniqueness of this case lies in the rarity of the site of this disease leading to a difficulty in diagnosis along with the difficulty in its management due to its locally recurrent nature and potential for distant spread.

Case Report

A 25-year-old female presented to the outpatient department with complaints of a painful swelling over her left lower jaw for the last 2 months. There was also associated history of loss of appetite and loosening of teeth above the swelling. On clinical examination, there was a hard, nontender swelling, around 5 cm × 4 cm in size involving the left hemimandible. It was extending anteriorly from the lateral incisors to posteriorly up to the second molar. It seemed to be infiltrating the lower alveolus and the gingivobuccal sulcus. The patient had no trismus or ankyloglossia, and the rest of the oral cavity did not present any abnormal findings.

A magnetic resonance imaging scan of the face and neck showed a 4-cm × 3-cm × 5-cm expansile lesion over the ramus of the left half of the mandible which was hypointense on T2-weighted images [Figure 1]. A whole body ^{99m}Tc-methylene diphosphonate scan picked up a primary lesion in the left mandible without any skeletal metastases [Figure 2]. The biopsy of the swelling revealed atypical round-to-oval cells with high nuclear: cytoplasmic ratio arranged haphazardly in a network of interconnecting variable-sized malignant osteoid tissue, suggestive of a high-grade osteogenic sarcoma [Figure 3]. A computed tomography (CT) scan of the chest showed multiple subcentrimetric ground-glass nodules in the right upper and middle lobes which were difficult to characterize.

With this clinicoradiological picture, the patient was treated with neoadjuvant

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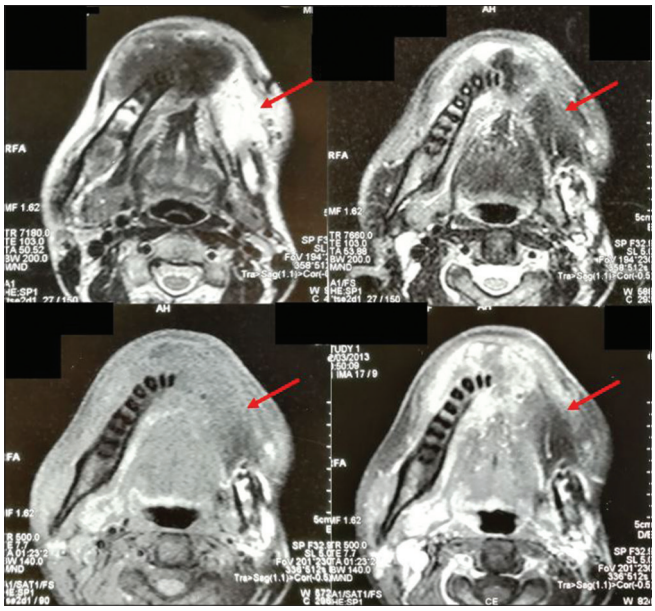

Figure 1: Magnetic resonance imaging image showing expansile lesion affecting the left hemimandible (red arrow)

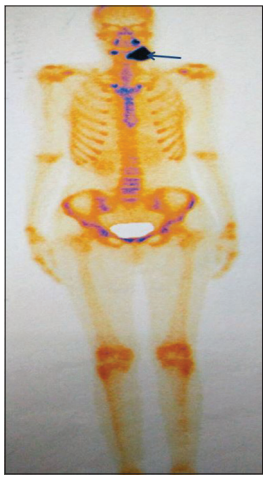

Figure 2: Whole body ^{99m}Tc-methylene diphosphonate scan showing a primary lesion in the mandible (blue arrow)

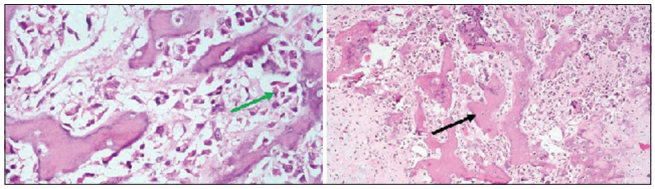

Figure 3: Biopsy of the lesion showing atypical round-to-oval cells (green arrow) with high nuclear: Cytoplasmic ratio arranged haphazardly in a network of interconnecting variable-sized malignant osteoid tissue (black arrow), suggestive of a high-grade osteogenic sarcoma

chemotherapy, with 4 cycles of ifosfamide, adriamycin, and cisplatin. On completion of chemotherapy, a response assessment imaging was carried out in the form of a CT scan of the face and chest. This was suggestive of a partial reduction in the tumor size in the mandible and complete disappearance of all nodules in the lung. The case was discussed in the multimodality tumor board, and in view of the patient's young age and the excellent response of her disease to chemotherapy, it was decided to treat her with a curative intent. She underwent a wide local excision of the tumor with a left hemimandibulectomy and a free osteomyocutaneous fibular flap. The postoperative histology showed clear margins with good response to chemotherapy (<10% of the tumor showed viable tissue). This was followed by two more cycles of adjuvant chemotherapy with ifosfamide and etoposide. She was kept on a three monthly follow-up and remained disease-free for the next 3 years. In the 4th year, she developed a local recurrence, for which she underwent a re-excision. Post the second surgery, she was planned for further chemotherapy but was found to have developed multiple bony and lung metastasis. She received palliative radiotherapy for painful bony metastasis to the left hemipelvis to a dose of 20 Gy in five fractions and is now on palliative chemotherapy with docetaxel, gemcitabine, and bisphosphonates. She has achieved an overall survival of 5 years since her diagnosis and presently has metastatic but stable disease.

Discussion

Osteogenic sarcoma of the mandible is a rare malignant tumor of the bone. The mandible presents a rare site of involvement with a distinct clinical profile with an older age of presentation and lesser chances of distant metastasis. However, Garrington *et al.* reported distant metastases in approximately 50% of the cases. The rate of local recurrences also seems to be higher leading to increased mortality.[7,8] Clinically, OS of the jaw bones tends to present commonly as a swelling with associated paresthesia which is dissimilar from the presentation of the disease of long bones where pain during activity is the more common complaint.[3,9]

Several factors predispose to the development of OS with rapid bone growth figuring as a prominent one. It has associations with other bone diseases, such as Paget's disease, fibrous dysplasia, enchondromatosis, and hereditary multiple exostosis. There is now a known association of OS with retinoblastoma involving the RB1 gene.[10]

Pathologically, high-grade intramedullary OS is the classic or conventional form comprising nearly 80% of the tumors.[11] Low-grade intramedullary OS constitutes <2%, and the other pathological types include parosteal and periosteal OS, which has juxtacortical or surface variants, accounting for <5%. Consistent with the above distribution, majority of mandibular OSs are conventional. OS spreads microscopically through the narrow spaces between bone tissue with other possible routes being the mandibular canal and the structures which connect the intraosseous components and soft tissues, such as the periodontal ligament and the mental

and inferior alveolar nerves. These structures may facilitate the extraosseous spread of an intraosseous lesion which can also occur through a recently extracted tooth socket or by perforation of the cortical plates.[10] Route of distant metastasis is hematogenous with the most common site being the lungs.

The radiographic picture of OS ranges from osteolytic to mixed to osteogenic pattern of bone. The widening of periodontal ligament space and inferior dental canal, together with sunburst effect are almost pathognomonic of OS of the jaw bone, but not all lesions may show these characteristics.[12]

OS of the jaw has a clinically distinct profile from that of the long bones although early and prompt diagnosis with interplay of surgery and chemotherapy remain the cornerstone of management.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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