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Gopal Chandra Halder, Santanu Patsa, Riteshkumar Baldevbhai Jadav, Jay Gopal Ray

ABSTRACT

Introduction: Osteosarcoma is common primary malignancy of bone that arises from the mesenchymal cells. It is commonly seen in younger patients having average age of 15 years. Generally, it involves long bones with fastest growth rate. The exact etiology for this tumor remains to be unknown but in some cases it runs in families. Osteosarcoma involving the jaw bones is relatively less frequently seen. Generally, osteosarcomas have diverse radiological and histopathological appearances.

Case Report: A 31-year-old male presented with a small asymptomatic gingival growth in the lower right posterior region for a period of 20 days. The lesion extended both on the buccal and lingual aspect of 46 and 47 regions.

Conclusion: The subtle and asymptomatic clinical and radiological features of this type of lesion as seen in the present case may delay early diagnosis and poor treatment outcome. The purpose of this study was to report an osteosarcoma involving the lower jaw which presented as a soft tissue mass, mimicking an inflammatory gingival lesion.

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CASE REPORT

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Introduction: Osteosarcoma is common primary malignancy of bone that arises from the mesenchymal cells. It is commonly seen in younger patients having average age of 15 years. Generally, it involves long bones with fastest growth rate. The exact etiology for this tumor remains to be unknown but in some cases it runs in families. Osteosarcoma involving the jaw bones is relatively less frequently seen. Generally, osteosarcomas have diverse radiological and histopathological appearances. **Case Report:** A 31-year-old male presented with a small asymptomatic gingival growth in the lower right posterior region for a period of 20 days. The lesion extended both on the buccal and lingual aspect of 46 and 47 regions. **Conclusion:** The subtle and asymptomatic clinical and radiological features of this type of lesion as seen in the present case may delay early diagnosis and poor treatment outcome. The purpose of this study was to report an osteosarcoma involving the lower jaw which presented as a soft tissue mass, mimicking an inflammatory gingival lesion.

Keywords: Asymptomatic osteosarcoma, Mandible, Orthopantomogram (OPG), Soft tissue

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INTRODUCTION

Osteosarcoma is the second most common primary bone tumor after multiple myeloma. The tumor usually arises from the metaphyseal growth plate of the long bones of the extremities of which 50% occur from the knee [1]. Osteosarcoma can be primary or secondary. Primary osteosarcoma can be classified into three sub types: intramedullary, surface and extraskeletal [2]. Some histopathologists classified the tumor into the following types: osteoblastic, chondroblastic and fibroblastic depending on the amounts of osteoid, cartilage and collagen fibers presenting in the examined tissue section [3]. Occurrence of the tumor in the jaws is about 6–9% of the all osteosarcomas [4]. The incidence is slightly higher in blacks than in whites (Huvos et al. 1983). Male to female ratio is 1.5:1 and taller patients are more commonly affected in compared to normal of same age group [5]. Osteosarcoma is very rare in young children (<5 years). Primary osteosarcoma typically occurs in young patients (10–20 years) among which 75% occurs before the age of 20 years. The reason behind is that the growth centres of the bone are more active in puberty to adolescence time period [1]. Secondary osteosarcoma occurs in elderly patient, usually secondary to Paget's disease or post radiotherapy period. The mean age of mandibular osteosarcoma is 34–36 years and it is often considered as a distinct entity because of its predilection to older patients [1].

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Jaw osteosarcoma is a rare and aggressive type of malignant tumor which is usually found in the third and fourth decades of life. Both jaws are involved with equal frequency without any gender predilection but in case of long bones, it has slightly higher frequency in male than in female [6]. It is frequently found at the posterior part of body and ramus of mandible. The maxillary tumours develop from the alveolar ridge, the sinus floor and palate. Pain and swelling are most common sign and symptom associated with looseness of teeth, lack of healing at the extracted site, hypoesthesia or paraesthesia in case of mandibular tumor [7, 8]. The biological behavior of the osteosarcoma in the long bones differ with the jaws osteosarcoma. There have been few reports of metastasis from jaw Osteosarcoma (1% of all malignancy) to a distant parts of body as compared to osteosarcoma of long bones which makes jaw osteosarcomas prognostically better [7, 9]. This case report emphasizes on the fact that asymptomatic lesions may occur in the jaws which can create diagnostic dilemma and delay early diagnosis.

Treatment of the jaws osteosarcoma is not well understood but in the case of long bones, it is well established. Disease free survival rate has been increased (from 20% in 1960s to 70% in 1980s) in the long bones osteosarcoma with the adjuvant of chemotherapy but it is not established in jaws osteosarcoma. Local recurrence is still a major cause of death in jaws osteosarcoma [8].

CASE REPORT

A 31-year-old male reported with a soft tissue swelling at right posterior lower molar regions for 20 days. Patient's chief complaint was only swelling without any pain or discomfort, looseness of teeth, bleeding from the lesion or paresthesia of lips. Patient gave a history of extraction of the lower right second molar about 6–7 years back due to caries. The past medical history was not-significant. Extraoral examination did not reveal any facial asymmetry or palpable lymph nodes on either right or left submandibular and sublingual regions at the first reporting time in the outpatient department of Dr. R. Ahmed Dental College and Hospital. Intraoral examination revealed a diffuse erythematous soft 1.2x0.8 cm swelling in the 46 and 47 regions which appeared like a localized gingival granulation tissue mass. Surface epithelium was ulcerated occlusally and covered with slough. The presumptive diagnosis was an inflammatory gingival lesion on the basis of clinical findings. Orthopantomogram showed a well circumscribed approximately 0.5x0.5 cm diameter radiolucent area at the distal root apex of 46 without any root resorption (Figure 1). Distal root surface of 46 and mesial root surface of 48 showed widening of periodontal ligament space (Figure 1). A routine hemogram was advised. After obtaining written consent from the patient and his relative, incisional biopsy procedure was performed from the site of lesion under local anesthesia in the department

of oral pathology. The tissue was preserved in 10% neutral buffered formalin and sent to the histopathology laboratory of Dr. R. Ahmed Dental College and Hospital, Kolkata for further processing. The tissue was embedded in paraffin wax after fixation and processing to prepare the wax block. Three serial sections from the wax block were made with the hand operated microtome (Leica Model No.RM2125RTS) in 4 μ m thickness. The lesion kept on growing very fast in size (Figure 2).

Histopathological examination of the H&E stained sections revealed a parakeratinized stratified squamous epithelium with elongated rete ridges along with the fibrocellular connective tissue. The deeper part of sections showed round to spindle-shaped pleomorphic cells with hyperchromatic and bizarre nuclei which were arranged in an irregular pattern (Figure 3). Some parts of the sections revealed atypical eosinophilic osteoid cells (Figure 3). In addition to osteoid, the tumor cells produced chondroid material and fibrous connective tissue (Figure 4 and Figure 5). Histopathological features were suggestive of osteoblastic osteosarcoma. After final diagnosis, the patient was referred to the Chittaranjan Cancer Hospital, Kolkata, West Bengal. Patient was advised for computed tomography (CT) scan of the head and neck, chest and abdomen to exclude secondary metastasis. Image analysis revealed no secondary metastatic lesions. However, images and other reports of these investigations were not shared with us due to hospital rules. The lesion kept on growing very fast in size (Figure 2). Radical hemimandibulectomy followed by chemotherapy was done (Figure 6). In the regimen of chemotherapy patient was given doxorubicin (25 mg/m²) by intravenous route on first, second and third days with Cisplatin only on first day (100 mg/m²). Cycle was repeated after 21 days interval and six such cycles were given.

DISCUSSION

Primary osteosarcoma in jaws is a rare lesion. Early metastasis of osteosarcoma in lung is common but metastasis to jaw bones is extremely rare, accounting

Figure 1: Orthopantomogram showing 0.5x0.5 cm radiolucent area at root apex of mandibular right first molar.

Figure 2: Showing a firm, sessile tumor like growth in right mandibular alveolar ridge extending up to buccal vestibule, two weeks after incisional biopsy.

Figure 5: Formation of osteoid and pleomorphic and hyperchromatic cells in connective tissue stroma (H&E stain, x100).

Figure 3: Neoplastic osteoid and tumor cells (H&E stain, x400).

Figure 4: Osteoid formation (H&E stain, x100).

for 1% of all malignancy [3]. Histopathologically, osteosarcoma is categorized into three subtypes—osteoblastic, chondroblastic and fibroblastic among which osteoblastic subtype is found in 60% of cases [4]. The classification was made depending on the relative amount of osteoid, cartilage, or collagen fibers production by the tumor cells. Sometimes, histopathological differentiation of osteosarcoma from malignant histiocytomas may be difficult [10]. Osteosarcoma predominantly occurs in rapidly growing bones [1]. Some chemical agents such as methylcholanthrene and chromium salt are linked to osteosarcoma [11]. The p53 and retinoblastoma (Rb) genes are well known as a tumor suppressor genes. This genes may become mutated, resulting in loss of normal protective function of body against a developing tumor. Mutations in both p53 and Rb genes have been found in the pathogenesis of osteosarcoma [12]. The p53 gene is mutated in 50% of all cancers and 22% of osteosarcoma [13]. Microscopic spreading of osteosarcoma is facilitated through narrow space. An intra-osseous lesion can spread to the adjacent tissue through periodontal ligaments, inferior alveolar canal, mental canal or through recently extracted tooth socket. It is often difficult to diagnose the lesion early due to its variable clinical and radiological features. Histopathological examination is the gold standard for final diagnosis. Immunohistochemical analysis was not done in this case. Literatures revealed that tumor cells showed positive staining for CD99, MIB-1 and S-100 and negative for AE1, AE3 and SMA [6]. Asymptomatic nature as seen in this case may delay histopathological examination. This case report can help to throw light on such lesions.

Osteosarcoma of jaw is an aggressive neoplasm with high rate of mortality despite a relatively low risk of distant metastases [14]. Early diagnosis and radical surgery followed by radiotherapy and/or chemotherapy have been found to result in good prognosis. Local recurrence after surgery is a major problem. Uncontrolled growth of

Figure 6: Postoperative view after hemimandibulectomy.

jaw osteosarcoma is a major cause of death for patients than are the effect of distant metastases. Most common sites of metastases are lungs and brain. Metastases is not frequent from mandibular lesion with respect to maxilla but local recurrence is more frequent in maxillary region due to its anatomical closeness to other bones. In this case, distant metastasis was not found prior to surgery. Patient also withstood the complete course of chemotherapy and is still in good health.

CONCLUSION

A dental surgeon is in a unique position to diagnose very early oral mucosal changes associated with such aggressive lesions since intraoral examination is a consistent part of dental treatment procedures. Early diagnosis of an asymptomatic lesion is quite difficult due to the consciousness of patients. Such asymptomatic early lesions must be kept in mind during intraoral examination. This article may help to the dental professionals during the intra oral examination which may guide them to diagnose the lesion at an early stage, result in better prognosis.

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Author Contributions

Gopal Chandra Halder – Conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Critical revision of the article, Final approval of the version to be published

Santanu Patsa – Acquisition of data, Drafting the article, Critical revision of the article, Final approval of the version to be published

Riteshkumar Baldevbhai Jadav – Acquisition of data, Drafting the article, Critical revision of the article, Final approval of the version to be published

Jay Gopal Ray – Acquisition of data, Drafting the article, Critical revision of the article, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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