CASE REPORT

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Periodontal manifestation of osteosarcoma of the fibula: a case report and review of literature

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Abstract

Osteosarcoma is a rare and aggressive malignant mesenchymal tumor. It usually presents in the long bones about a decade prior to that in jaws. This clinical condition is characterized by radiolucent to radiopaque masses, sunburst appearance, codman's triangle and periodontal ligament space widening in the jaws. A male aged thirty years reported to us with chief complaint of severe pain in gums in the lower left back region. Intraoral examination revealed tobacco stains on his teeth revealing his habit of tobacco chewing and bidi smoking. There was moderate amount of plaque deposition and periodontal pocket ranged from 2 to 5 mm in depth. Orthopantogram revealed widening of periodontal ligament space in the 35, 36, 37, 38 region. The patient also gave history of severe bone pain in left leg for last 3 years and was referred to orthopedic department where he was diagnosed as a case of osteosarcoma of left fibula after various hematological and radiological investigations. Clinical acumen in detecting unusual periodontal ligament widening with no apparent cause as in this case lead to detection of osteosarcoma in the left fibula after clinical, radiological and histopathological correlation emphasizing the role of inter disciplinary approach.

Key words

Fibula, histopathology, ligament, mandible, orthopantanogram, osteosarcoma

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Background

Osteosarcoma is an aggressive mesenchymal tumor considered as the most common primary bone malignancy, characterized by formation of osteoid like tissue in the lesion [1, 2]. It usually presents in the long bone about a decade prior to that in jaws [3]. Manifestation of osteosarcoma of jaw is a painful condition with symptoms like swelling of bone and adjacent soft tissue, tooth bulging, dislocation *etc.* Other features include lack of healing and swelling at the site of the tooth extraction, trismus and hypoesthaesia or paresthaesia in the case of the mandibular tumours, nasal obstruction in the maxilla [4]. Symmetric widening of the periodontal ligament space of the involved teeth termed as positive Garrington's sign, also noticible in some cases [5]. Knowledge and understanding of radiological features of osteosarcoma can help in earlier diagnosis, shortens delay in treatment and will ultimately favour the prognosis.

Osteosarcomas are broadly divided into two major categories - primary and secondary. Primary types are further classified as intramedullry/central and surface osteosarcomas as per guidelines of World Health Organization classification [6].

Table – 1 Types of Osteosarcomas

Primary Osteosarcomas	Secondary Osteosarcomas
Conventional-intramedullary/central high grade(commonest)	Secondary Osteosarcomas may occur as a result of radiation exposure, mostly seen in Paget's disease[6, 7]
a.Osteoblastic (50%)	
b.Chondroblastic (25%)	Osteoblastic osteosarcoma-sclerosing type
c. Fibroblastic (25%)	Osteosarcoma resembling osteoblastoma
Small cell	Chondromyxoid fbroma-like osteosarcoma
Telangiectatic	Chondroblastoma-like osteosarcoma
Low grade central	Clear-cell osteoblastoma
Surface osteosarcomas:	Malignant fibrous hisiocytoma-like
Parosteal	osteosarcoma
Periosteal	Giant cell rich osteosarcoma
High grade surface	Epithelioid osteosarcoma

Osteoblastic type is characterized by varied shapes and size in neoplastic osteoblasts with large deeply staining nuclei, arranged in a disorderly fashion, accountable for majority (60%) of the jaw lesion. Head and neck region is more prone for chondroblastic type of Osteosarcomas with distinguishable features like atypical chondroid areas which were composed of pleomorphic cells with large hyperchromatic nuclei and prominent nucleoli [8]. Fibroblastic type is relatively uncommon and noticed in the jaws. Most of the tumours are heterogenous, which is an indicative of pleuripotency of the proliferating mesenchymal cells [9, 10]. The radiographic appearance can be varied ranging from radiolucent to radio opaque masses, moth eaten appearance, codman's traiangle and periodontal widening [11].

Exact cause of osteosarcoma is still under research, but ample scientific reports pointed out defects in RB and p53 genes leads an important role in this pathophysilogical process. Germline mutations in RB increases the risk of osteosarcoma 1000-fold. It has also observed that patients suffering from Li-Fraumeni syndrome (germline p53 mutation) are vulnerable for this tumor [7].

The rate of incidence of osteosarcoma for all races and both genders are 4.0 (3.5-4.6) for the range 0-14 years and 5.0 (4.6-5.6) for the range 0-19 years per year per million persons [2].

This report described a case of periodontal manifestation of osteosarcoma of the fibula, which clinically presented as gingival recession, necrotizing ulcerative periodontitis lesion, moderate alveolar bone loss, widening of periodontal ligament space and osteolytic lesion at the proximal end of the left fibula. This report is also important because it highlights its early diagnosis based on simple, yet rare radiographic and histologic findings despite its misleading clinical presentation with osteomyelitis with proliferative periostitis, suppurative osteomyelitis, ossifying fibroma and osteoblastoma.

Case Report

A male patient aged 30 years age referred to Dr. Ziauddin Ahmad Dental College & Hospital, Aligarh Muslim University, Faculty of Medicine, Dept. of Periodontics & Community Dentistry, Aligarh, Uttar Pradesh, India, with chief complaint of severe pain in gums with dry mouth.

General examination was normal and patient appeared well oriented to time and place. The patient complained of severe pain in the gums in lower left back region. On oral examination there were severe tobacco stains on all teeth of patient which revealed to us that he was on habit of eating pan-masala and bidi smoking for the last 6-7 years [Figure1].

Figure 1 - Showing severe and generalized tobacco stains on the teeth of the patient.

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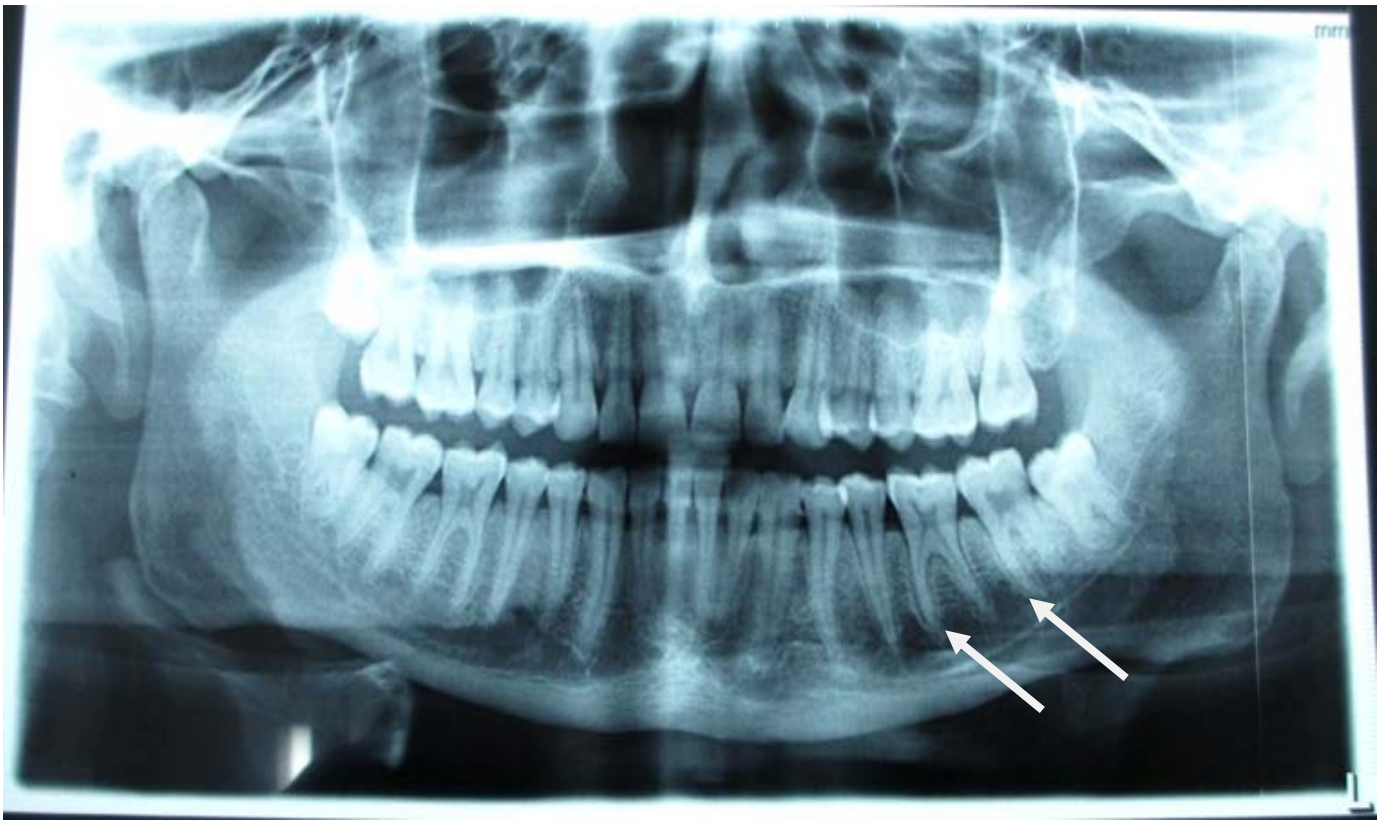

Figure 2 - Orthopantogram (OPG) of the patient showing periodontal ligament space widening around 35,36,37 & 38 reminiscent of osteosarcoma.

The oral mucosa was blanched and pale and there were gingival recession present in relation to 31, 41 with necrotizing ulcerative periodontitis lesion in relation to these teeth. On periodontal examination periodontal pocket ranged from 2 to 5mms and the plaque deposition was moderate.

Orthopantanogram (OPG) of the patient revealed moderate alveolar bone loss and the most remarkable feature was widening of periodontal ligament space with respect to 35, 36, 37, 38 [Figure 2] reminiscent of oral manifestation of osteosarcoma in the jaw. Patient was further questioned regarding any other physical illness or complaint. Patient revealed severe pain in the left leg which had been present for past 3 years. Patient was referred to orthopedics department for the same where after co-relation of radiological and histopathological investigations [Figures 3 &4].

The patient was diagnosed as a case of osteosarcoma of left fibula. Clinical acumen in detecting unusual periodontal ligament widening with no apparent cause as in this case lead to detection of osteosarcoma in fibula.

Discussion

Osteosarcoma of jaws is a relatively infrequent condition with overall prevalence of 8% of all osteosarcoma [10]. Osteosarcoma is notorious condition which is usually diagnosed in advanced stages. After being confirmed by the radiographic features of affected bone, histopathological examination was performed to diagnose osteosarcoma.

Influence of gender

In this report, the subject was male. This is in accordance with other well documented literatures, where male predominance was reported (5.4 per million persons per year in males vs. 4.0 per million in females) [2]. Another research showed the male: female ratio 1.2:1. Longer period of skeletal growth and additional volume of bone in men, may be a reason for this, still more research is required to confirm it [12].

In this case we found tobacco stains on all teeth of patient an indicative of pan-masala and bidi smoking for few years. This was also an important risk factor for the development and severity of inflammatory periodontal disease [13]. Our patient complained severe pain in gums with dry mouth. A study by clark *et al*. on 66 patients, showed almost similar complaints [10].

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Figure 3 - Radiograph of the left leg showing osteolytic lesion at the proximal end of the left fibula.

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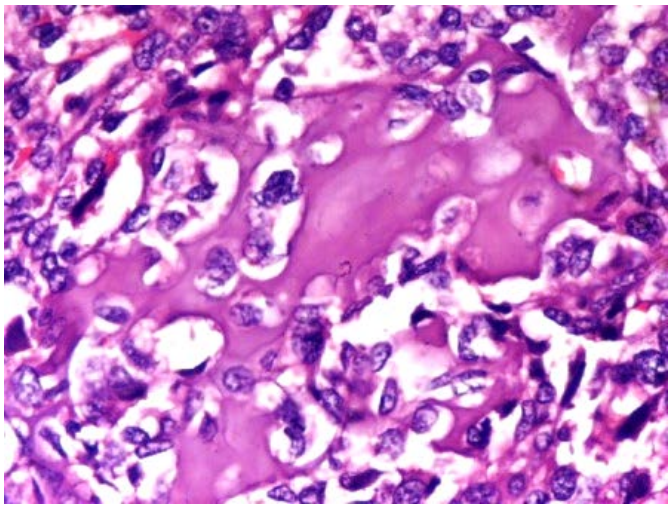

Figure 4 - Photomicrograph shows highly pleomorphic spindle shaped cells with scattered bizarre cells with osteoid production. Hematoxylin and Eosin x 40.

Clinical manifestation in Osteosarcoma

Although ossification in the soft tissue component of the bone manifesting as a "sunburst" pattern is a distinctive feature in this case, not considered for specificity. In some cases of the osteosarcoma of the jaw Periosteal new bone formation occurs with lifting of the cortex which ultimately forms Codman's triangle [14-17]. Although earlier reports mentioned that roentgenographic evidence of a symmetrically widened periodontal membrane space is a remarkable feature for early diagnosis in osteosarcoma of the jaw, but same pattern of changes observed in some cases of chondrosarcomas [8].

In this case diagnosis of extraoral (left fibula) osteosarcoma manifestation in the mandible was made because of the striking radiographic finding of periodontal widening. Epidemiological scenario clarifies most frequent sites of osteosarcoma are metaphyseal growth plates of long bones like femur (42%, with 75% of tumors in the distal femur), the tibia (19%, with 80% of tumors in the proximal tibia), and the humerus (10%, with 90% of tumors in the proximal humerus) [2]. Due to similarity with other clinical conditions like osteomyelitis with proliferative periostitis, suppurative osteomyelitis, ossifying fibroma, osteoblastoma, sometimes there is delay in diagnosis of Osteosarcomas [18]. Dental professional may often be the first to notice such an aggressive lesion. In most of the cases of osteosarcoma, the lesion would have already advanced by the time it is established. Hence, to recognize the unusual feature in oral radiographs for example periodontal widening and co-relating it clinically may prevent undue morbidity or mortality.

This is a case report of unusual presentation of periodontal widening which lead us to suspect and to identify osteosarcoma in fibula.

Conclusion

This case report highlights the importance of recognizing atypical findings in oral cavity. Radiographic finding of abnormal periodontal widening suggested underlying pathology lead to diagnoses of this potentially fatal condition. In conclusion, Osteosarcoma if diagnosed early can save patient from unwarranted morbidity. Hence, triad of rapid diagnosis, multi-disciplinary approach towards diagnosis, management and long term observation is the key to manage osteosarcoma.

Abbreviations

Orthopantanogram (OPG), tumor protein 53(p53)

Competing interests

None.

Authors' contribution

Dr. Saif khan took part in the concept, review of literature, manuscript drafting, case work-up and follow-up. Dr. kafil akhtar also contributed in the review of literature, drafting, case work-up and follow-up. Dr. Pritma Singh helped in the case-work-up and follow-up, drafting of the manuscript.

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