Case report

PAROSTEAL OSTEOSARCOMA OF THE MAXILLA: A case report and review of the literature

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ABSTRACT

OBJECTIVE: A report of a rare case of parosteal osteosarcoma, a low-grade malignant lesion diagnosed in our Centre, and to highlight the diagnostic challenges, treatment considerations and prognosis of the lesion; compared to other reactive, benign and malignant jaw lesions.

CASE REPORT: A 67 years old female who presented with a slow growing oval shaped, well circumscribed, bony hard, left maxillary swelling of 4 months duration; extending from the region of the left upper lateral incisor tooth (22) to the left upper first molar tooth (26) and fixed to the overlying gingiva. Oblique lateral radiograph showed the left maxillary labial alveolus with lobulated ossified mass. The clinical differential diagnoses of the lesion were osteoma and fibrous dysplasia. Histopathological diagnosis of parosteal osteosarcoma was made after incisional biopsy of the lesion. Surgical resection (with wide margin) of the affected maxillary segment and the associated teeth was performed and post-surgical biopsy confirmed the diagnosis of parosteal osteosarcoma. No recurrence was observed after 4 years follow up of the patient.

CONCLUSION: Parosteal osteosarcoma shares similar clinico-pathological characteristics with some periosteal lesions. However, special imaging techniques and histopathological evaluation remains the most reliable tools for definitive diagnosis of these lesions. This study recommends close cooperation between Oral and Maxillofacial Surgeons, Pathologists and Radiologists in the management of parosteal osteosarcoma.

Key words: Parosteal osteosarcoma, Maxilla, diagnostic-challenge, treatment, prognosis

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INTRODUCTION

Parosteal osteosarcoma is a low-grade malignant lesion accounting for 1.6 to 2% of all malignant bone tumours. The lesion is found commonly in the distal, dorsal femur, with a wide age range of 2nd to 8th decades of life and female predilection. [1,2]In contrast, recent report by Samardzisk et al,[1] shows a slight male predilection for parosteal osteosarcoma, which presented mostly as painless swelling with average duration of presentation of 12.5 months. Radiologically the lesions showed a densely ossified and lobulated mass. The histology of most of the lesions showed regular trabeculae interspersed with a scarcely cellular, spindle shaped cell and collagenous stroma, with a low to moderate cellular atypia and low mitotic activity.

However, parosteal osteosarcoma is rare in the orofacial region and most cases reported were found in the maxilla, occurring in both genders. [3-5] Hewitt et al reported [3] a case of maxillary parosteal osteosarcoma of 1 year duration in a 32 years old male that presented as a firm, painless maxillary swelling with palatal extension. The patient had a partial Maxillectomy without adjunct therapy and no evidence of local recurrence was observed after 3 years. Bianchi et al [5] reported a maxillary parosteal osteosarcoma with a sclerotic radiological pattern. Also, radiographically the typical appearance is that of an exophytic mass which has a lobulated configuration and is adjacent to bone with central dense ossification.[6]A fine radiolucent cleft may be found between the tumour and adjacent outer table in cranial parosteal osteosarcoma. [7] The string sign has been described as representing the cleavage plane separating the tumour and adjacent normal cortex.There is often cortical thickening and lack of aggressive periosteal reaction.[6] This lesion radiologically mimicks myositis ossificans[8] and half of the cases of myositis ossificans adheres to the periosteum and are referred to as parosteal myositis ossificans.[9,10] Other parosteal lesions that are commonly misdiagnosed as parosteal osteosarcoma, because they share similar clinicopathological characteristics with parosteal osteosarcoma are osteochondroma,[1,2]osteoma, juxta-cortical chondrosarcoma, high-grade surface osteosarcoma, and periosteal chondroma.[11-14]

This study reports a rare case of parosteal osteosarcoma, a low-grade malignant lesion diagnosed in our Centre, and to highlight the diagnostic challenges, treatment considerations and prognosis of the lesion; compared to other reactive, benign and malignant jaw lesions.

CASE REPORT

A 67 years old female patient presented with a slow growing left maxillary swelling of 4 months duration, in the Oral and Maxillofacial Surgery Department, University of Benin Teaching Hospital, Benin City, Nigeria. The lesion was oval shaped, well circumscribed, bony hard, measuring 5cm by 2cm, extending from the region of the left upper lateral incisor tooth (22) to the left upper first molar tooth (26) and fixed to the overlying gingiva [Figure 1].

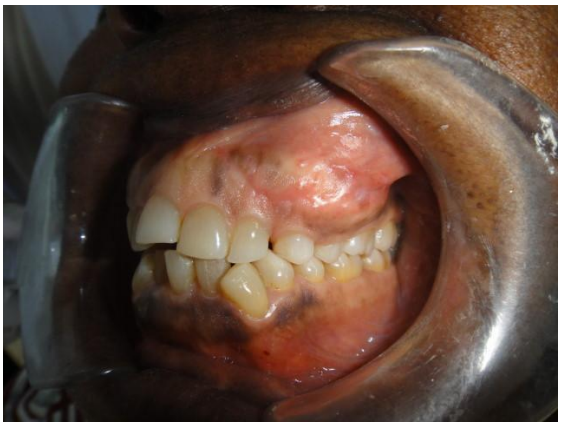

Figure 1: Clinical photograph showing parosteal osteosarcoma presenting as a left maxillary swelling.

Plain radiographs including posterior-anterior skull view, oblique lateral and occlusal views of the maxilla were taken. Only the oblique lateral view clearly demonstrated the lesion in the left maxillary labial alveolus with lobulated ossified mass. The surrounding alveolar bone and adjacent dentition appeared normal. [Figure 2].

The clinical differential diagnoses of the lesion were osteoma and fibrous dysplasia.

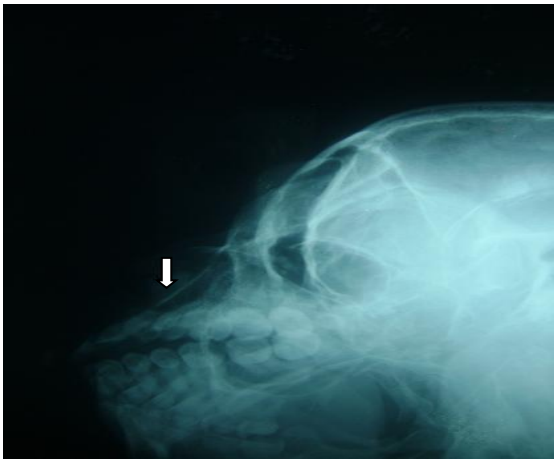

Figure 2: Oblique lateral radiograph showing the left maxillary labial alveolus with lobulated calcific mass.

Histopathological diagnosis of parosteal osteosarcoma was made after incisional biopsy of the lesion. The lesion showed thick islands and streaming parallel trabeculae of woven bone with intervening sparsely cellular fibrous connective tissue stroma, and outer thick periosteum (fibrovascular capsule) [Figure 3]. There are areas showing low cellular atypia consisting of bone cells with pale, pleomorphic nuclei and no obvious mitosis [Figure 4].

Surgical resection (with wide margin) of the affected maxillary segment and the associated teeth was carried out and post-surgical biopsy confirmed the diagnosis of parosteal osteosarcoma. No adjunct therapy was included in the patient's treatment. The patient attended 2 postoperative follow-up visits in 1 month, with operation site showing defect in the left maxilla (Figure 5). The patient was referred to the prosthodontist for review and rehabilitation with upper partial denture to replace the missing teeth. Further follow-up visits in the Oral and Maxillofacial Surgery Unit were scheduled for 3 months, 6 months and yearly period. There was no evidence of post-operative complication or recurrence after 4 years of follow up.

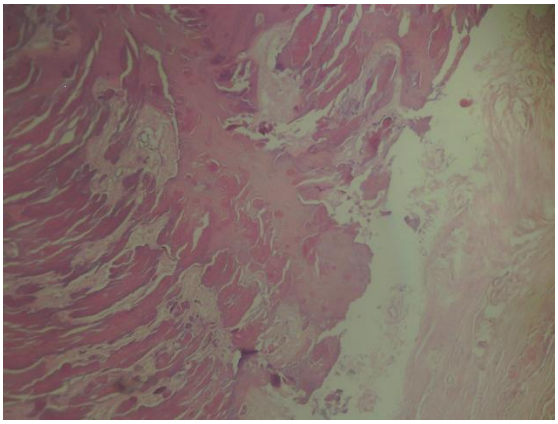

Figure 3: Photomicrograph showing parosteal osteosarcoma with streaming parallel trabeculae of woven bone with intervening sparsely cellular fibrous connective tissue stroma and outer periosteum [H&E x100]

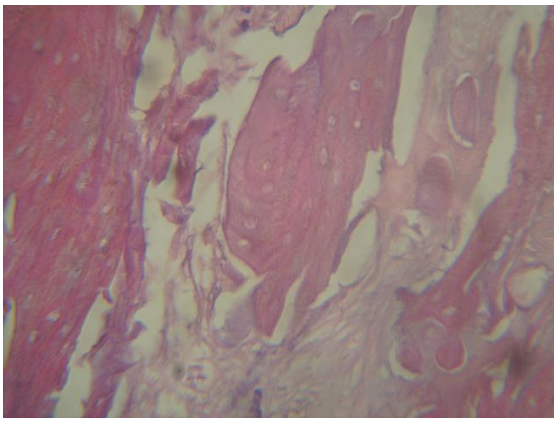

Figure 4: Photomicrograph showing parosteal osteosarcoma with low cellular atypia consisting of bone cells with pale, pleomorphic nuclei and no obvious mitosis [H&E x400]

DISCUSSION

Parosteal osteosarcoma is low grade malignant lesion found rarely in the orofacial region, with male predilection and wide age range on presentation of the lesion. The lesion may be misdiagnosed as a reactive or benign lesion because of the less aggressive clinical course and the excellent prognosis.[3-5]

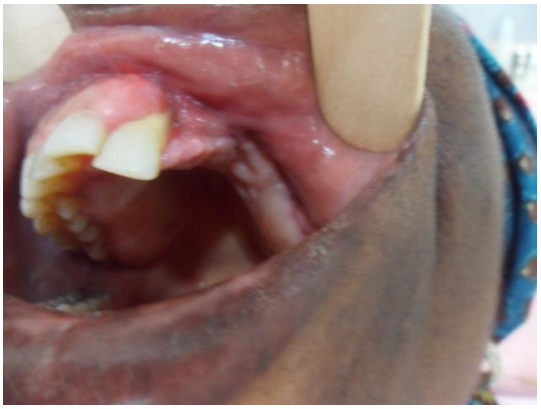

Figure 5: Clinical photograph showing post-surgical left maxillary defect

However, this study reports a case of parosteal osteosarcoma in an elderly female patient, with a long standing history of maxillary swelling, which was clinically misdiagnosed as osteoma or fibrous dysplasia. This is consistent with previous reports of misdiagnosis of parosteal osteosarcoma for other bone forming tumours including osteoma and fibrous dysplasia.[11,15] Cross-sectional imaging with computed tomography and magnetic resonance imaging, although not done in the case presented, gives better anatomic delineation of such lesions and would have assisted in definitive diagnosis of this lesion.[16] The recognition of the distinct radiological and histopathological characteristics of these lesions are useful to ascertain the definitive diagnoses of these lesions.[11] The diagnostic challenges and treatment considerations for parosteal osteosarcoma emphasizes the need for close cooperation between Oral and Maxillofacial Surgeons, Pathologists and Radiologists in the management of this lesion.

A comparison of the treatment and prognosis of low-grade parosteal osteosarcoma with those of the suspected reactive lesion (fibrous dysplasia) and benign lesion (osteoma) shows that parosteal osteosarcoma requires wide margin of resection to prevent recurrence, [3,13,14] while fibrous dysplasia is usually a self-limiting lesion, which may require chemotherapy (with non-steroidal anti-inflammatory drug [NSAIDs] and bisphosphonate) or surgical paring down; and it has potential for malignant change.[17] Similarly, osteoid osteoma has recently been reported to have potential to regress, so conservative medical treatment with NSAIDs is recommended, while surgery is reserved for rare cases with persistent symptoms. [18]

Previous reports [2,19] on the pathomorphology of surface osteosarcoma identified the histological variants of this lesion as low-grade parosteal osteosarcoma (Broders grade 1 to 2), periosteal osteosarcoma, an intermediate lesion (Broders grade 2 to 3), high-grade surface osteosarcoma (Broders grade 3 to 4), and dedifferentiated parosteal osteosarcoma, a high-grade lesion (Broders grade 3 to 4). Bertoni et al, [19] also observed that surgical resection with wide margin, without adjunct therapy was adequate for low-grade parosteal osteosarcoma and this gives good prognosis; whereas the dedifferentiated parosteal osteosarcoma required surgical resection with wide margin, together with adjunct chemotherapy, to avoid metastasis of the lesion and a high rate of mortality.In agreement with the prescribed treatment options for histological diagnosis of a low-grade parosteal osteosarcoma. [19] The patient in this study had only surgical resection (with wide margin) without adjunct therapy. Also, rehabilitation of the postoperative maxillary defect with upper partial denture was initiated within the 1 month follow-up visits. Similar to previous report, there was no recurrence of the lesion after 4 years of follow up.[3]

In conclusion, parosteal osteosarcoma shares similar clinico-pathological characteristics with other periosteal lesions. However, special imaging techniques and histopathological evaluation remains the most reliable tools for definitive diagnosis of these lesions. This study recommends close cooperation between Oral and Maxillofacial Surgeons, Pathologists and Radiologists in the management of parosteal osteosarcoma.

Conflict of Interest: None declared

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