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RESEARCH ARTICLE

PLEXIFORM AMELOBLASTOMA INVOLVING THE MANDIBLE IN AN ELEVEN YEARS OLD CHILD: A UNIQUE CASE REPORT

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ABSTRACT

Ameloblastoma is the most common benign but locally aggressive odontogenic neoplasm affecting the jaws, accounting for 1% of all tumors of the maxilla & mandible and 11% of all odontogenic tumors. Though nearly half of the tumors do occur between the ages of 20 and 40 years, it is relatively rare in paediatric patients and accounts for approximately 10-15% of all reported cases. Recurrence frequently appears after inadequate treatment. A case of plexiform ameloblastoma in a eleven years old male child with it's clinical, radiological, histopathological features and treatment modalities have been discussed herewith.

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INTRODUCTION

Ameloblastoma is a benign but locally aggressive, true epithelial odontogenic neoplasm that may arises from the dental lamina, developing enamel organ, dental follicle, periodontal ligament, lining of odontogenic cyst (dentigerous cyst) or even may arise from basal cells of oral mucosa (Gorlin, 1961; Appel, 1985 and Stafne, 1975). It represents about 1% of all oral ectodermal tumour and 9 % of all odontogenic tumors (Adebiyi, 2006). Ameloblastoma usually found in mandible (80%) (Sehdev, 1974). 70% of neoplasm are usually located in molar and ascending ramus region, 20% in the premolar region and 10% in anterior region of jaw bone, primarily seen 3rd to 5th decade of life without any definite sex predilection (Adebiyi, 2006 and Sehdev, 1974). Neoplasm is exclusively rare in children (represent 10 to 15% among all reported cases in paediatric age group) (Keszler, 1986). Clinically it usually appears as a slow enlarging, asymptomatic swelling causes expansion and distortion of the cortical plates and displacement of regional teeth leading to obvious facial asymmetry. Rarely, it can ulcerate through the mucosa (Nakamura, 2001). Radiologically ameloblastoma reveal a unilocular or multilocular radiolucency produces a typical honeycomb or soap bubble type of appearance along with thinning of cortical plates. Impaction of regional tooth may be noted occasionally.

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Radiologically it may be mistaken for Odontogenic keratocyst, Dentigerous cyst, Aneurysmal bone cyst and Giant cell tumour etc (Tozaki, 2001 and Gümgüm, 2005). Histologically different varieties of ameloblastoma are found namely follicular, plexiform, acanthomatous, granular, desmoplastic basal cells, clear cell variants and more rarely kerato-ameloblastoma and papilopherous ameloblastoma. Neoplasm is characterized by neoplastic ameloblast like cells showing the presence of hyperchromatic nucleus situated away from the basement membrane (reverse polarity) and the intervening connective tissue stroma is relatively loose vascular with minimum cellular components10. Surgical excision is the main stay of treatment and the recurrence rate usually ranges from 55 to 90% but the incidence of recurrence may decrease up to 5 to 10 % following radical resection (Gardner, 1984; Dolan, 1981 and Muller, 1985). In this article, we present a unique case of plexiform ameloblastoma of an eleven years old male child involving the mandible with brief clinical, radiological and histopathological features.

Case Report

An eleven years old male child from semi urban area reported to the Department of Oral & Maxillofacial Pathology, Guru Nanak Institute of Dental Sciences and Research, Panihati, Kolkata with a chief complain of swelling along with mild pain and discomfort involving the left side of lower jaw for the last three months.

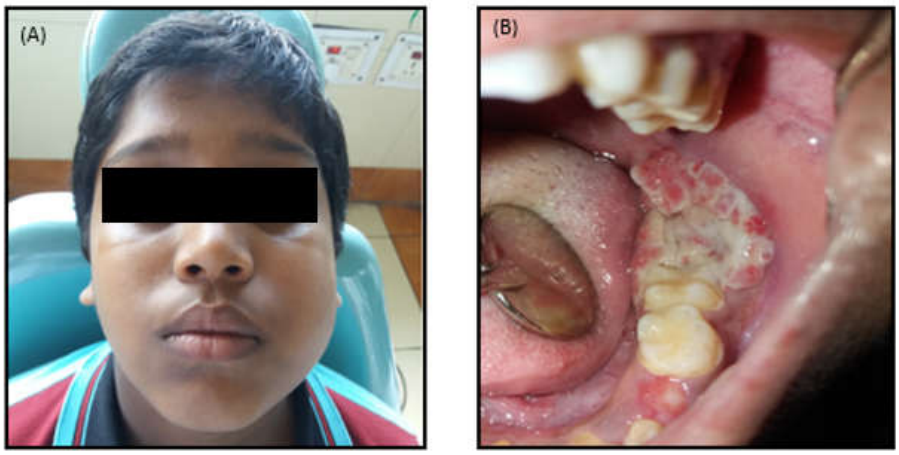

Figure 1 (A) Extra oral photograph shows swelling on left lower third of face: (B) Intra oral photograph shows exophytic, ulcerated lesion involving 36 region associated with expansion of buccal cortical plate.

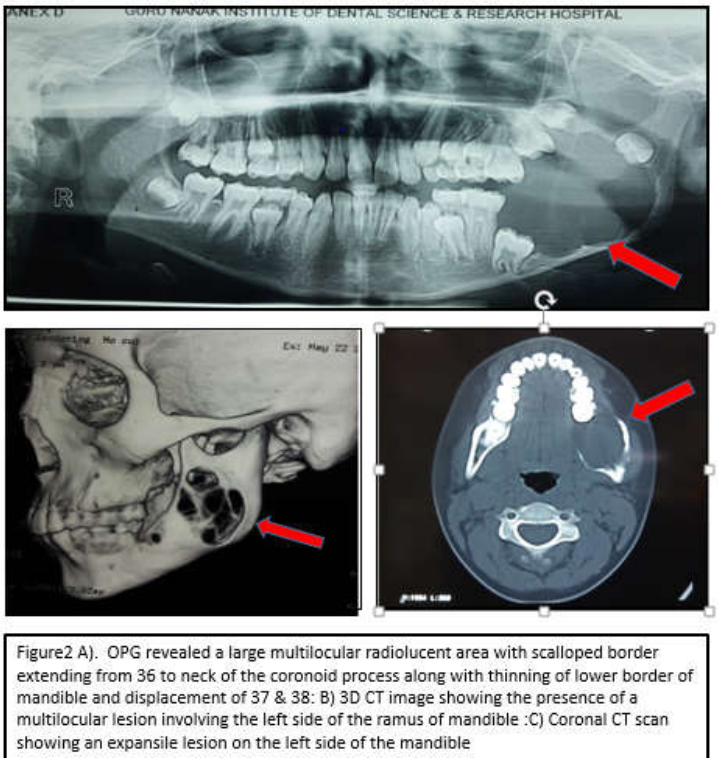

Figure2 A). OPG revealed a large multilocular radiolucent area with scalloped border extending from 36 to neck of the coronoid process along with thinning of lower border of mandible and displacement of 37 & 38: B) 3D CT image showing the presence of a multilocular lesion involving the left side of the ramus of mandible :C) Coronal CT scan showing an expansile lesion on the left side of the mandible

The lesion was initially small and asymptomatic which gradually enlarges and attain the present size over the time. On extra oral examination there was a well-defined uniform swelling involving the middle and lower third of the face adjacent to the body and ramus extending from the left zygomatic region to the lower border of mandible. The regional ipsilateral lymph nodes were tender, palpable and overlying skin was free. Intra oral examination revealed a diffuse ovoid intraosseous swelling measuring about 3x5 cm extending from 36 to retromolar region along with ulcerated overlying surface (Figure 1). On palpation the lesions was bony hard , associated with cortical plate expansion, being accompanied by egg shell crackling especially on lingual side and regional tooth were mobile. Routine haematological and biochemical investigations were within normal limit. Orthopantomogram (OPG) revealed a large multilocular radiolucency involving left side of body of mandible extended from 36 to the neck of the coronoid process along with thinning of lower border of mandible and inferior displacement of 37. Developing 38 was pushed towards the condylar process .CT scan showed an expansile lytic lesion in the posterior aspect of left side of mandible with cortical thinning and perforation of both buccal and lingual plate (Figure 2). Based upon the clinical and radiological findings, our provisional diagnosis was in favour of odontogenic cysts or neoplasm. An incisional biopsy was performed from representative site of the lesion under local anaesthesia and the specimen was processed for histopathological examination. Histopathology revealed the presence of long anastomosing cords of neoplastic odontogenic epithelium with peripheral tall columnar cells exhibiting reverse polarity resembling ameloblasts. The central cells were loosely arranged mimicking stellate reticulum .The supporting connective tissue stroma showed moderate vascularity and chronic inflammatory cells infiltration (Figure 3). The histopathological diagnosis was made as Plexiform Ameloblastoma. The patient was referred to the Dept. of Oral& Maxillofacial surgery for further treatment & management.

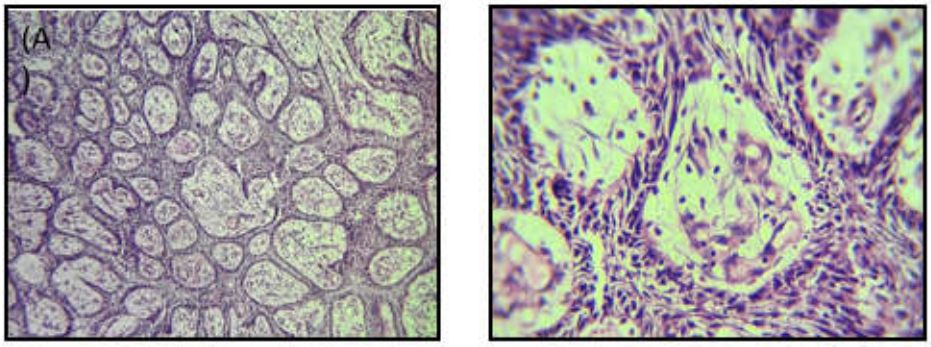

Figure 3 (A) Low power (H&E, 10X) shows anastomosing cords of odontogenic epithelial cells enclosing connective tissue stroma. (B) High (H&E, 40X) shows odontogenic epithelium with peripheral tall columnar cells exhibiting reverse polarity resembling ameloblasts. The central cells were loosely arranged mimicking stellate reticulum.

DISCUSSION

Ameloblastoma is a benign but locally aggressive true odontogenic neoplasm with capacity to attain great size, erode bone and invade adjacent structure13. Safer et al14, postulated that ameloblastomas may arises from either cell rest of enamel organ, epithelium of odontogenic cyst, developing enamel organ or basal cells of surface epithelium. The term ameloblastoma was coined by Churchill in 1933 but first detailed description of the lesion was established by Falkson in 187913. It is the most common odontogenic tumor, although it represents only about 1% of tumors and cysts of the jaws (Nakamura, 2001). The age distribution of ameloblastomas is from 20 to 50 years 15-17 without definite sex predilection although higher frequency in female has been described predominantly in molar premolar region (Iordanidis, 1999). In the present case a large ameloblastoma was found in an eleven years old male patient in molar and ascending ramus region and associated with unerupted teeth but the patient was eleven years male which is relatively uncommon. The case under discussion revealed a large diffuse swelling involving left body and ascending ramus of the mandible leading to mild facial deformity.

The swelling was hard, mild tender showing bucco-lingual cortical plate expansion and covered by ulcerated surface. Egg shell crackling on lingual side and mild paresthesia were noted all of which mimic the conventional clinical features as discussed in previous literatures (Nakamura, 2001 and Torres-Lagares, 2005). Radiologically it revealed a large well defined multi-locular radiolucency involving the left side of the body of mandible in molar premolar area. CT scan showed expansile, lytic lesion in posterior aspect of left side of mandible with cortical plate expansion, perforation. This radiological findings were in accordance with the previously published literature (Gümgüm, 2005 and Kim, 2001). Histopathologically ameloblastoma is characterized by the proliferation of odontogenic epithelial cells arranged on a stroma of conjunctive vascular tissue in locally invading structure which resemble the enamel organ at diverse stage of differentiation (Kim, 2001). Various histological pattern have been described in the previous literatures and include follicular, plexiform, acanthomatous, desmoplastic, papiloferous. The tumour found in our patient was an ameloblastoma of plexiform type having anastomosing cords with peripheral tall columnar ameloblast like cells exhibiting hyperchromatic nuclei situated away from the basement membrane mimicking reverse polarity along with centrally located scanty stellate reticulam like cell as depicted in the previous case series (Nakamura, 2001; Kim, 2001 and Kovács, 1999). Treatment of ameloblastoma depends on age, general health, size, location and duration of the tumour and is controversial in children due to continued facial growth of different bone, presence of un-erupted teeth and presence of greater percentage of cancellous bone. The treatment varies from conservative approach to radical resection. The conservative approach includes enucleation followed by chemical cauterization, marsupialization and segmental resection (Shafer, 1983; Regezi, 1978; Guerrisi, 2007). Hong et al (2007), in a retrospective analysis of 239 patients with Ameloblastomas of the jaws reported the recurrence of the disease is about 4.5% in patient treated segmental resection and maxillectomy, 11.6% which were treated by resection with bone margin, and 29.3% treated with conservative treatment. Scariest et al (Scariot, 2012) reported that there was no recurrence in 9year old child having unicystictic ameloblastomas who was treated with curettage and extraction of two premolar mandibular teeth. In conclusion we emphasize that late presentation of tumors is a frequent problem in low and middle income countries. Early diagnosis, conservative timely intervention and conservative surgical treatment should be the first choice for treating ameloblastomas in children and it will be imperative to further refine our understanding of disease both clinically and molecularly to improve the precision with which we treat ameloblastomas.

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