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PLEXIFORM AMELOBLASTOMA OF MANDIBLE: A CASE REPORT

Dental Science

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ABSTRACT

Ameloblastoma is a benign, locally aggressive odontogenic tumor originating from the odontogenic epithelium. It appears most commonly in the third to fifth decades and with equal frequency between sexes. It manifests as a slow-growing swelling, causing expansion of the jaw bones. Ameloblastoma prevalently occurs in the mandibular molar and the ramus areas. Radiologically, it presents as a unilocular or multilocular radiolucent lesion characteristic of honeycomb or soap bubble appearance. Ameloblastoma reveals several histologic patterns in which foremost histopathological patterns are the follicular and plexiform types, followed by the acanthomatous and granular cell types. Recurrence frequently appears after inadequate treatment. We present the extensive case of plexiform ameloblastoma of the mandible with its clinical, radiological, histological features and treatment modalities.

KEYWORDS

Plexiform ameloblastoma, Mandible, Odontogenic tumor

INTRODUCTION

Ameloblastoma is the most common of all odontogenic tumors. It is a true neoplasm of the odontogenic epithelium. It is a benign and locally aggressive neoplasm of odontogenic epithelium that arises from the remnants of the dental lamina and odontogenic epithelium. Ameloblastoma appears most commonly in the third to fifth decades but the lesion can be found in any age group including children. Plexiform ameloblastoma is one of the variants of ameloblastoma, with specific histopathological features.[1-4]

A tumor is usually asymptomatic and presents itself as a slowly enlarging facial swelling. It is characterized by slow growth and local infiltration into the adjacent tissues. It is an aggressive tumor, 70 to 80% of ameloblastomas develop in the molar-ramus region of the mandible. Radiographically an ameloblastoma can be a unilocular or multilocular radiolucent lesion with a honeycomb or soap bubble appearance.[1-3,5,6]

Ameloblastomas comprise only 1 % of all oral tumors. (WHO)'s classification of head and neck tumors stated four forms of ameloblastomas: 1. multicystic, 2. Unicystic, 3. peripheral, and 4. desmoplastic ameloblastomas. Multicystic ameloblastoma is common and represents 86% of all the cases. The chief histopathological types of ameloblastoma are the follicular and plexiform, followed by the acanthomatous and granular cell types. The plexiform pattern is less aggressive and has a significantly lower recurrence rate.[1,2,4]

The aim of this case report is to present a clinical case of an ameloblastoma which radiographically differentiated with other lesions such as Central giant cell granuloma, Keratocystic odontogenic tumor, Odontogenic myxoma, and Aneurysmal bone cyst, and surgically lesion was enucleated, and histopathologic diagnosis suggested plexiform ameloblastoma.

Case Report

A 55-year-old female patient reported to oral medicine and radiology department, with a chief complaint of swelling over the lower part of the face causing facial asymmetry for the last 6 months. The patient was asymptomatic 6 months earlier she noticed a painless swelling over the left side of the face. The patient realized the swelling gradually increase in size so she visited at a civil hospital at Surat where case history recorded, X rays & FNAC were done which suggestive of odontogenic tumor. So they referred to the civil hospital in Ahmedabad for further diagnosis and treatment plan. She had a history of snuff 3 to 4 times since 20 years.

On extraoral examination facial asymmetry present. A well defines swelling size approximately 5*3 cm extending from right side of parasymphysis region to left side of the angle of the mandible and superoinferiorly from the corner of mouth to the inferior border of the mandible. The skin over swelling was normal in color and texture. No any bleeding or pus discharge. The swelling was nontender, hard in consistency, smooth, with well-defined margins [Figure: 1].

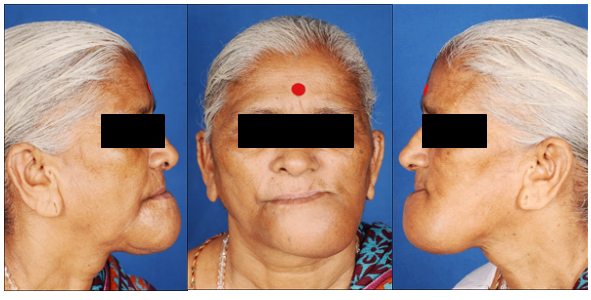
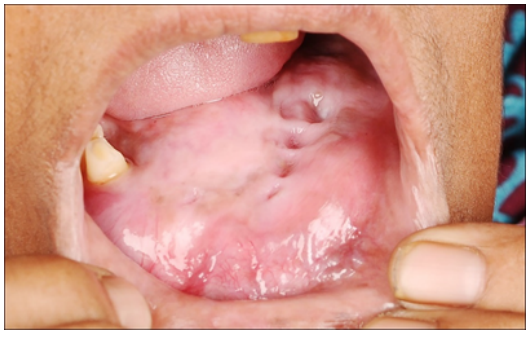


Figure: 1: front and side view of the patient]

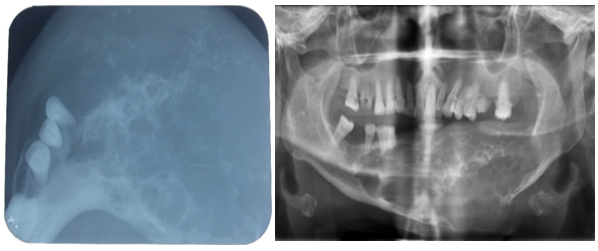
Intraoral examination showed well define swelling size approximately 7*5 cm extending from right mandibular 2nd premolar to left retromandibular pad area and 5 cm buccal and lingual cortical plate expansion. Normal overlying mucosa and upper teeth indentation present over the swelling. 46 and 42 to 37 teeth were missing. The swelling was nontender, firm to hard in consistency, egg shell crackling present [Figure: 2].



[Figure: 2: Clinical intra oral picture]

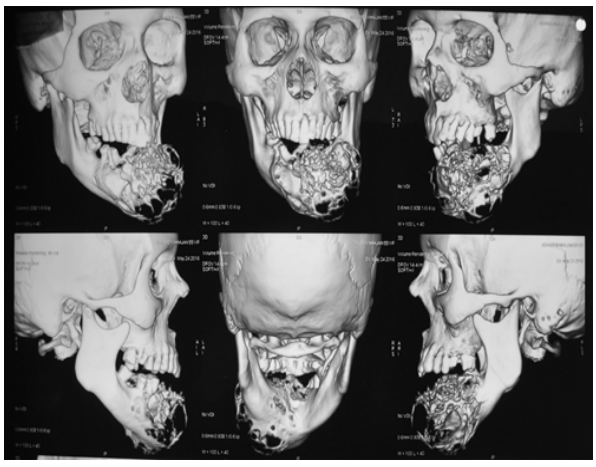
The mandibular cross-sectional occlusal radiograph showed well define multilocular lesion with ill-defined thin curved septa with buccal as well as lingual cortical expansion with a break in continuity associated with displacement of 43, 44, and 45 [Figure: 3].

Panoramic radiograph (OPG) showed a single well define multilocular radiolucent lesion size approximately 7*4 cm extending from mesial aspect of 45 to body of the left side of the mandible at 38 regions and superoinferiorly from alveolar crest to the inferior border of the mandible. The internal structure was multilocular with ill define thin curved septa with soap bubble and honeycomb appearance with blunt root resorption of 43, 44 and 45 with thinning and expansion of inferior border of the mandible with pathological fracture present associated with generalized horizontal bone loss [Figure: 3].



[Figure: 3: Mandibular cross-sectional occlusal and OPG radiographs]

CT scan showed large expansile multiloculated lytic lesion involving the central arch and bilateral body of the mandible. Lesion shows a presence of multiple internal calcifications and hyperdense solid component. Also evidence of anterior cortical break as well as few posterior cortical breaks and scalloping [Figure: 4].

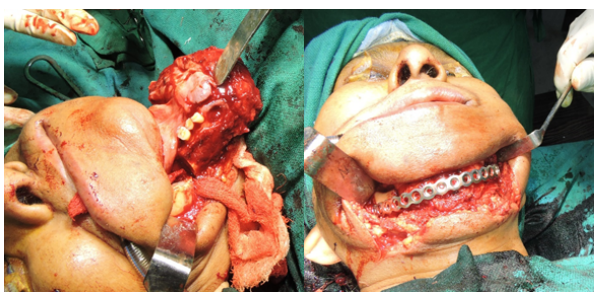


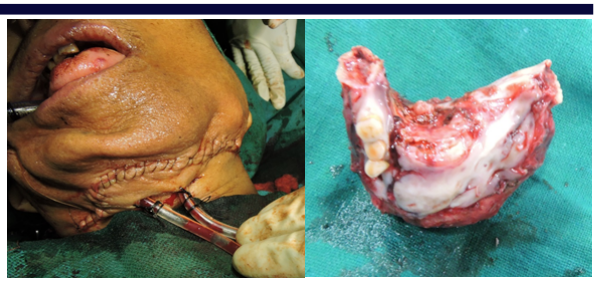
[Figure: 4: CT scan]

Based on the above findings our probable diagnosis was central giant cell granuloma. While ameloblastoma, keratocystic odontogenic tumor, calcifying epithelial odontogenic tumor, and odontogenic myxoma kept as a differential diagnosis.

Incisional biopsy of the lesion was done and histological examination revealed a calcifying epithelial odontogenic tumor (pindborg tumor). H & E stained sections showed proliferating tumor epithelial component in the form of strands, discrete islands, sheets of polyhedral epithelial cells and fibrous stroma. There was a presence of numerous amorphous eosinophilic strands and islands.

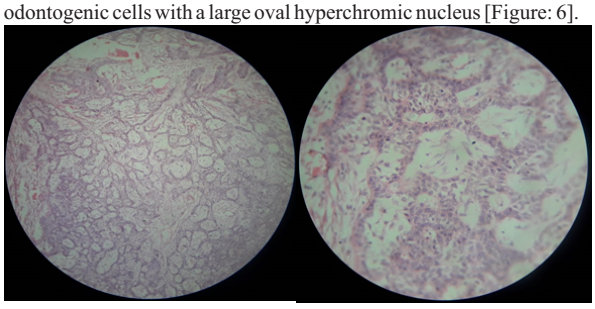
On the basis of biopsy, under a general anesthesia segmental mandibulectomy was done taking safe margins of 1 cm [Figure: 5]. Reconstructed was done with AO titanium reconstruction plate and iliac crest graft. No recurrence was observed during the 1 years follow up period.



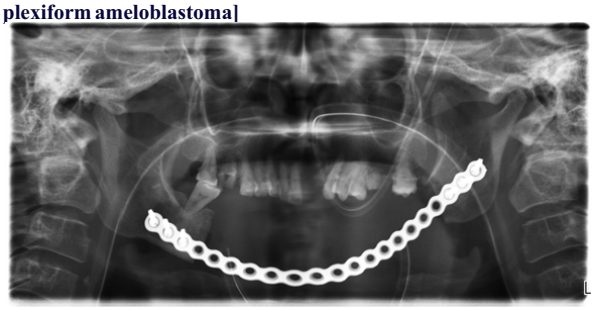


[Figure: 5: Intra operative procedure and specimen pictures]

Biopsy specimen's histopathological finding was suggested of plexiform ameloblastoma. H & E stained section showed a fibrocellular connective tissue mass interspersed with ameloblastic follicles, areas resembling plexiform ameloblastoma and sheets of odontogenic cells with a large oval hyperchromic nucleus [Figure: 6].



[Figure: 6: Histopathological picture of the tumor revealed a plexiform ameloblastoma]



[Figure: 7: Postoperative OPG]

DISCUSSION

The term ameloblastoma was coined by Churchill in 1933, adamantinomas having been previously reported by Falkson and Malassez in 1879. It is the most common odontogenic tumor although it represents only about 1% of tumors and cysts of the jaws. In the mandible, 70% are located in the molars or the ascending ramus areas, 20% in the premolar region, and 10% in the anterior region of the mandible. About 10-15% of ameloblastomas are associated with an unerupted tooth. In the present case, a large plexiform ameloblastoma found in the anterior and molar region of the mandible. [2,4,5,7]

Ameloblastoma appears equal frequency in both the sexes, although a higher frequency in females than in males has been described. In our case, the patient was female and was in the 6th decade of her life. [2,5,6]

Clinically, it appears as a painless swelling, which can be associated with gradually increase facial asymmetry, a teeth in the involved region may be displaced and become mobile, malocclusion, ulceration, and paresthesia of the affected area may present. In our case, clinical examination revealed a large, expansive mass in lower anterior and left a posterior molar region of the mandible. The swelling was hard, painless to palpation and covered by normal mucosa with upper teeth indentation present over the swelling. [5,6]

In most cases, ameloblastoma presents a characteristic but not diagnostic radiographic appearance. Radiographically, ameloblastoma may present in different patterns. The most common pattern is a multilocular appearance with various cysts that are in groups or separated by osseous reinforced septa (soap bubble appearance). Another form is the unilocular form. Resorption of the adjacent tooth roots is not uncommon. In many cases, an unerupted

tooth, most often a mandibular third molar, is associated with the tumor. Sometimes ameloblastoma is indistinguishable from a dentigerous cyst. Ameloblastoma has a persistent and slow growth, spreading into marrow spaces with pseudopods without concomitant resorption of the trabecular bone. As a result, the margins of the tumor are not clearly evident radiographically or grossly during operation, and the lesion frequently recurs after inadequate surgical removal. In our case, it was a multilocular appearance with ill define thin curved septa with soap bubble and honeycomb appearance associated with blunt root resorption of the teeth. [1,5]

In 1970, Vickers and Gorlin defined the histopathological features of ameloblastoma. These features include hyperchromatic, palisading and reverse polarization of the basal nuclei with vacuolization of the cytoplasm. [12] Histologically, it is characterized by the proliferation of epithelial cells arranged in a collagenous fibrous connective tissue stroma of conjunctive vascular tissue in locally invading structures that resemble the enamel organ at different stages of differentiation. The tumor found in our patient was a plexiform ameloblastoma. The term “plexiform” refers to the appearance of anastomosing islands of odontogenic epithelium in contrast to a follicular pattern. [2,5,6,8,9]

Ameloblastoma is usually resected En bloc and sometimes with hemimandibulectomy if the lesion is highly infiltrative and extensive. Surgical resection with margins of 1.2 cm has had the least rate of recurrence. In our case, surgical resection of large-sized ameloblastoma was done to prevent recurrence and mandible stabilized and reconstructed was done with AO titanium reconstruction plate and iliac crest graft [Figure: 7]. When treated inadequately, malignant development is a possibility which is 1.6 to 2%. Long-term follow-up is necessary because this lesion has been shown to recur 25 and 30 years following primary treatment. [5,6,7,8,10]

CONCLUSION

Ameloblastoma is considered as a benign locally invasive odontogenic tumor with a high rate of recurrence. Because of the tendency for this tumor to grow silently, it is usually quite advanced at the time of presentation. Resection with some safe margin is the best primary method for treating solid/multicystic ameloblastomas to avoid recurrence while curettage, local excision, radiotherapy, and chemotherapy may play a palliative role in treatment.

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