

## CENTRAL PLEXIFORM AMELOBLASTOMA WITH EXOPHYTIC GROWTH: A RARE CASE REPORT WITH REVIEW OF LITERATURE

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### ABSTRACT

*Ameloblastomas are predominantly benign, intra-osseous odontogenic tumors and mucosal involvement is a rare secondary phenomenon, occurring only after a long period of intra-osseous growth and bone expansion. This article presents a case report of an eleven year old male patient with a large plexiform ameloblastoma which invaded the soft tissues, presented as an exophytic growth in the mandibular anterior region with radiographic feature of a large unilocular radiolucency and displaced mandibular left central incisor. The management comprised of surgical removal of ameloblastoma under local anesthesia along with extraction of displaced mandibular left central incisor.*

**Key words:** Plexiform Ameloblastoma, Exophytic growth, Peripheral Ameloblastoma

### INTRODUCTION

Ameloblastomas may arise from rests of dental lamina, from a developing enamel organ, from the epithelial lining of an odontogenic cyst, or from the basal cells of the oral mucosa. They are slow growing, locally invasive tumors that run a benign course in most cases. They occur in three different clinicoradiographic patterns which deserve separate consideration because of differing therapeutic considerations and prognosis<sup>1-4</sup>. These are: conventional solid or multicystic (central; about 86% of all cases); unicystic (about 13% of all the cases); and peripheral (extraosseous; about 1% of all cases)<sup>1,2</sup>.

The central type ameloblastoma originates almost entirely from a central lesion of the jawbone and tends to expand the bone without perforating the jaw periosteum. Rarely this lesion penetrates the periosteum and extends into extraosseous soft tissues<sup>7,8,9</sup>.

The term plexiform unicystic ameloblastoma refers to a pattern of epithelial proliferation that has been

described in cystic lesions of the jaws. It does not exhibit the histological criteria for ameloblastoma published by Vickers and Gorlin<sup>5</sup>, and has therefore been considered by some pathologists to be a hyperplastic epithelial proliferation rather than an ameloblastoma. The peripheral ameloblastoma, also known as the extraosseous ameloblastoma, soft tissue ameloblastoma, ameloblastoma of mucosal origin, or ameloblastoma of the gingiva is a very uncommon odontogenic tumor<sup>1,2,4,6</sup> that accounts for 1% for all ameloblastomas<sup>1</sup>.

### CASE REPORT

An eleven year old male patient reported to the Department of Pediatric and Preventive Dentistry, with a complaint of swelling in the front region of the lower jaw which was present since a year. The present history revealed that the swelling was slowly growing along with a soft tissue growth inside

the oral cavity over a period of 3 months. The parents gave history of bleeding from the intraoral lesion on contact with any sharp food particles. The patient's medical history was not relevant, and his general health was good.

Extraoral examination revealed swelling in the left mandibular anterior region which extended from the mandibular midline to the corner of the mouth on the left side. The swelling was firm in consistency and was non tender on palpation.

Intraoral examination revealed a soft tissue swelling extending laterally from mesial surface of the mandibular left central incisor to the mesial surface of the mandibular left first primary molar, labially extending around 3cm obliterating the vestibule and extending into the floor of the mouth. The lesion was pebbly, sessile, firm and pink in color with ulcerated surface due to repeated masticatory trauma (Fig. 1). Bleeding from the lesion was seen on gentle probing. Grade II mobility was present with left central incisor.

The panoramic radiograph showed a large unilocular radiolucency extending from mesial surface of left central incisor to the mesial surface of first primary molar with the displacement of left central and lateral incisor (Fig. 2). The mandibular occlusal radiograph showed expansion of labial cortical plate with displaced left central incisor (Fig. 3).

Incisional biopsy was carried out and sent to the oral pathology department. The histopathological report was inconclusive apart from hyperplastic stratified squamous epithelium with fibrous tissue underneath. Hence complete enucleation of the lesion along with sub- periosteal dissection was planned under local anesthesia.

Mental nerve block along with local infiltration was given. Sulcular and relieving incisions were given using a no.15 BP blade. After reflecting the mucoperiosteal flap by periosteal elevator on both buccal and lingual side, the expanded cortical plate was identified and separated from the mucoperiosteum. Subperiosteal dissection was carried out beginning from the sound bone. The cystic lining was separated from the inferior border of the mandible and was surgically removed along

with displaced mandibular left central incisor (Fig. 4). The retracted cystic mass along with the incisor was then sent for histopathological analysis (Fig. 5). As the mucoperiosteal flap was not adequate for approximation and suturing, betadine guaze pack was placed in the cavity sutured using 3-0 silk suture materials to heal by secondary intention (Fig. 6). The size of betadine soaked guaze piece was progressively reduced every alternate day.

Microscopically, hematoxylin and eosin stained section showed numerous anastomosing and branching strands and cords lined peripherally by ameloblast-like cells. The strands and cords contained stellate reticulum-like cells within them. The intervening connective tissue was made up of loose fibrous stroma and showed many scattered blood vessels and hemorrhagic areas. (Fig.7a and 7b).

On the basis of clinical, radiographic and histopathological findings, the present case was diagnosed as plexiform ameloblastoma with exophytic growth. At the six month follow-up, healing was satisfactory without any sign of recurrence.

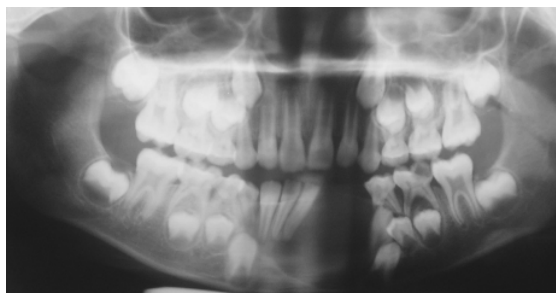
#### REVIEW OF LITERATURE

The term ameloblastoma was coined by Churchill<sup>10</sup> in 1934. As per Cakur et al.<sup>11</sup> the first detailed description of this lesion was given by Falkson in 1879<sup>12</sup>. Unicystic ameloblastoma was first described by Robinson and Martinez<sup>13</sup> in 1977. Peripheral ameloblastoma was first reported by Kuru<sup>14</sup> in 1911. In 1959, Stanley and Krogh<sup>15</sup> defined the clinical and histopathologic characteristics of peripheral ameloblastoma.

Ameloblastoma is uncommon in children. The most commonly quoted article regarding ameloblastoma is a review of 1,036 ameloblastomas, in which the average patient age is 38.9 years, with only 2.2% (19 of 858) under 10 years and 8.7% (75 of 858) between 10 and 19 years<sup>16</sup>. This report, however, was published in 1955, when adenoameloblastomas and ameloblastic fibromas were included under ameloblastomas and the histological characteristics for ameloblastomas in cysts had not been delineated.



**Fig. 1:** Intraoral view of the patient showing extensive lesion.



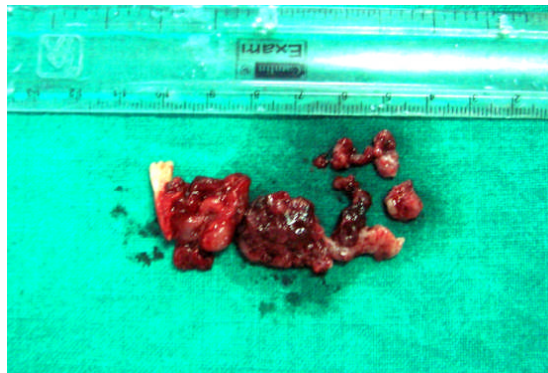
**Fig. 2:** Panoramic radiograph showing a unilocular radiolucency in the mandibular anterior region



**Fig. 3:** Mandibular occlusal radiograph showing expansion of buccal cortical plate and displacement of left central incisor.



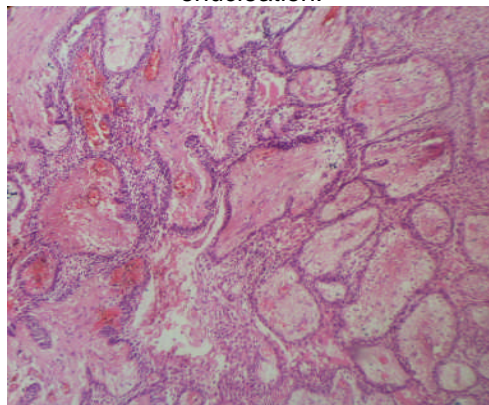
**Fig. 4:** Enucleation of the lesion along with removal of left central incisor



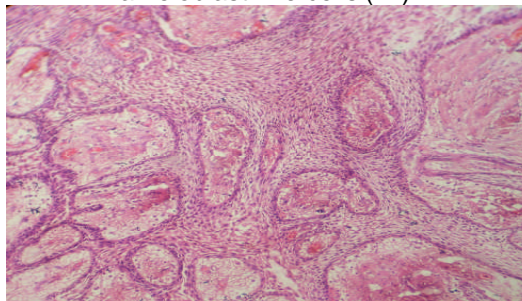
**Fig. 5:** Enucleated lesion along with the left central incisor



**Fig. 6:** Sutured site with betadine gauze pack after enucleation.



**Fig. 7a:** H & E stained section showing anastomosing cords and strands of ameloblast-like cells (4x).



**Fig. 7b:** H & E stained section showing ameloblast-like cells and stellate reticulum-like cells with hemorrhagic areas (10x).

However literature on peripheral ameloblastoma in children is scanty. Nauta et al.<sup>17</sup> presented 52 cases of peripheral ameloblastoma; cases of basal cell carcinoma of the gingiva and peripheral ameloblastoma. Various authors have reported the case of central ameloblastoma invading the soft tissues. Kawai et al.<sup>8</sup> reported a case of maxillary ameloblastoma in the form of polyp. Yoshikawa et al.<sup>9</sup> reported a case of ameloblastoma of the maxilla invading the palatal mucosal forming a ulcer. Gotoh et. al.<sup>18</sup> reported a case of ameloblastoma with ulceration on the distal gingiva of the mandibular second molar in a 29 year- old male patient. Stevenson and Austin<sup>19</sup> reported a case of ameloblastoma presenting as an exophytic gingival lesion in the left mandibular third molar region in a 47 year old female patient.

#### DISCUSSION

The ameloblastoma is a locally invasive neoplasm derived from odontogenic epithelium. This distinctive semimalignant, unicentric, nonfunctional, persistent odontogenic neoplasm shows intermittent or slow growth. The proliferating tumor may infiltrate the cancellous marrow spaces without causing bone destruction. It tends to expand the bone rather than perforate it. Occasionally patients allow an ameloblastoma to persist for many years without treatment and though the expansion may become extremely disfiguring the tumor does not break through the bone. But for reasons unknown some ameloblastomas manage to penetrate the bone and extend into the surrounding soft tissues<sup>20</sup>.

Philipsen et. al.<sup>6</sup> opined that several authors refer to Kuru<sup>14</sup> as having reported the peripheral ameloblastoma for the first time in 1911. However, what Kuru<sup>14</sup> described was not a peripheral but rather an intraosseous ameloblastoma having penetrated through the

alveolar bone, fused with the oral epithelium and eventually presenting itself clinically as a "peripheral lesion"<sup>21</sup>. Although the strict criteria of peripheral ameloblastoma might exclude an ameloblastoma with a recognizable intrabony lesion, several authors reported such a case as peripheral ameloblastoma<sup>22-24</sup>. The first completely documented case of a peripheral ameloblastoma must be attributed to Stanley and Krogh<sup>15</sup>, who defined the clinical and histopathologic characteristics in 1959.

In our patient, both panoramic and mandibular occlusal radiographs demonstrated bony destruction, though the findings were similar as that of Kuru<sup>14</sup> we diagnosed it as the central plexiform ameloblastoma with exophytic growth.

The differential diagnosis must be made with fibrous nodules, gingival tumours, peripheral odontogenic fibromas, peripheral ossifying fibromas, pyogenic granuloma, peripheral giant-cell granuloma and other peripheral hyperplastic swellings superficial to the alveolar ridge<sup>1-4,15</sup>. Simpson<sup>25</sup> reported that because of the location and histopathologic features it is possible to mistake peripheral ameloblastoma for basal cell carcinoma.

The histological diagnosis in this case presented a dilemma, in that lesion could be either be an intra-osseous ameloblastoma with a predominant mucosal component or a peripheral ameloblastoma causing bone resorption, both of which are rare manifestations. A detailed reappraisal of the clinical, radiographic and histological findings was needed before a definitive diagnosis was made.

The initial exophytic presentation of the lesion was suggestive of a peripheral ameloblastoma; however, the short history of exophytic growth, the presence of an underlying radiolucent lesion undermined the support for this diagnosis.

Radiographic feature of a large radiolucency and histological features of ameloblasts-like cells in plexiform pattern was seen. The tumor was, therefore, finally categorized as an intraosseous plexiform ameloblastoma extending to a peripheral site, merging with the overlying epithelium and then perforating, thus creating the exophytic lesion seen clinically.

The presented case is a rare entity because:

- a. The lesion was seen in a young patient of just 11 year old
- b. The lesion was located in the mandibular anterior region
- c. The lesion had penetrated the periosteum and breached through the overlying epithelium presenting as an exophytic growth which was considerably big in size.

### CONCLUSION

A rare case of an intraosseous plexiform ameloblastoma with interesting clinical, radiographic and histological findings has been presented with a review of literature. The presentation of the tumor as an exophytic lesion could invite diagnostic confusion with the more common exophytic lesions and the rare peripheral ameloblastoma. The importance of determining the pathogenesis of an ameloblastoma has been stressed so that the appropriate treatment can be provided.

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