Case Report

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Soft Tissue Recurrence of Plexiform Ameloblastoma in 8 years post Disarticulating Hemimandibulectomy: A Case Report

Abstract:
Ameloblastoma is characteristically considered to be a benign but locally aggressive and infiltrative odontogenic tumor with a high recurrence rate. Because of its locally invasive nature, the recurrence rate for the ameloblastoma post treatment remains high. Recurrence also seems to depend on the histological type of the tumor and the treatment modalities used. As per literature, curettage alone is reported to have a 90% recurrence rate for mandibular tumors and a 100%recurrence rate for maxillary tumors. Approximately 17.7% and 22.6% recurrence rates were also reported after radical and conservative treatment respectively; granular and follicular variants of ameloblastomas were seen to recur more frequently than the plexiform type. Most recurrences occur at the site of the primary tumor. However, lesions are also known to recur in bone grafts, and rarely, recurrence involves adjacent soft tissues. In this study, we present an unusual case of a plexiform ameloblastoma that recurred in the left mandibular residual soft tissue8 years after the first surgery i.e. disarticulating hemimandibulectomy. The patient is on regular follow up post second surgery for 23 months and she has been symptom free till now.

Key-words: Recurrent Ameloblastoma, Soft Tissue Recurrence, En-bloc Resection, Odontogenic tumors, Plexiform Ameloblastoma

Introduction:

Ameloblastoma is a true neoplasm of odontogenic epithelium[1] representing about 1% of all oral ectodermal tumors and 9% of odontogenic tumors[2]. It is an aggressive neoplasm which arises from the remnants of the dental lamina and dental organ. Most ameloblastomas develop in the posterior region of the mandible with 70% of these arising in the molar-ramus area and they are occasionally associated with the unerupted third molar teeth[3]. Ameloblastoma appears most commonly in third to fifth decades of life but the lesion can be found in any age group including children[4].

In a conventional radiograph, ameloblastoma can be presented as either unilocular or multilocular corticated radiolucency; the bony septae results in a honey comb or soap bubble appearance. In some places, cortical plates are spared and expanded where as in other region they are destroyed; root resorption is also a common radiological finding[5]. Conventional radiograph is sufficient for small mandibular lesions but maxillary lesions and extensive lesions require CT and MRI to establish the extent of the lesion[6].

The principal histopathological variants of ameloblastoma are the follicular and plexiform types, followed by acanthomatous and granular cell types. Uncommon variants include desmoplastic, basal cell type, clear cell ameloblastoma, keratoameloblastoma and papilliferous ameloblastoma[7].

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The challenge in managing ameloblastoma is in achieving complete excision and reconstruction of the defect when the tumour size is large. This tumor is treated by enucleation, curettage or surgical excision depending on size and the type of the lesion. The rate of recurrence ranges from 17.7% for en-bloc resection to 34.7% in case of conservative therapy. Wide resections with a safety margin of healthy bone to prevent local recurrence is still preferred[8].

Here, we report a case of recurrent plexiform ameloblastoma in residual alveolar mucosa of the left body region of mandible in a 27-year-old female 8 years after surgical resection and reconstruction using reconstruction plate.

Case Report:

A 27 year old female patient had reported to the Department of Oral and Maxillofacial Surgery with complaint of intraoral swelling on left side of her lower jawsince last 1 month. The patient underwent Disarticulating Hemimandibulectomy of left side with Reconstruction using titanium reconstruction plate [Fig. 1(a),1(b),1(c)] under general anaesthesia for histopathologically confirmed Plexiform ameloblastoma at the same location[8] years back. The patient also gave a history of extraction of left posterior mandibular molar teeth 2 month prior to the first surgery.

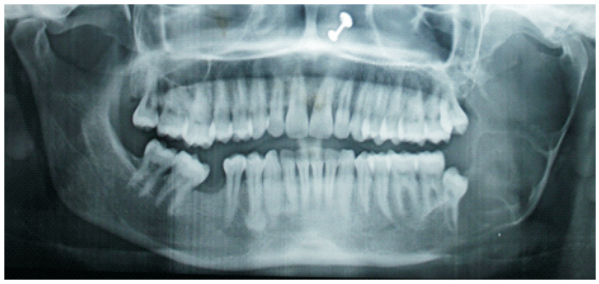

Fig 1(a): Pre-operative Orthopantomogram (OPG

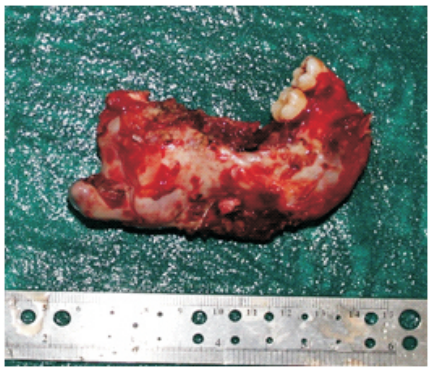

Fig 1(b): Resected Left Hemimandible with condyle

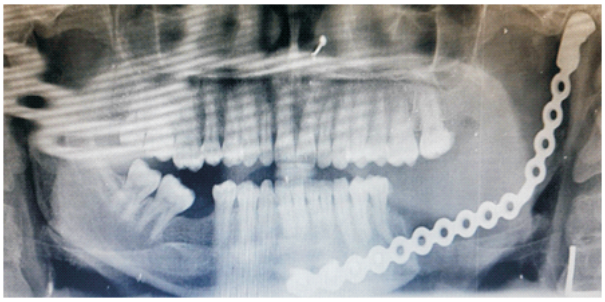

Fig 1(c): 3 months Post-operative Orthopantomogram (OPG)

Intraoral Clinical examination revealed non-peduncalated, smooth–surfaced, firm mobile swelling(size 2.5 cm × 1.5 cm)on left side of posterior residual alveolar mucosa***(Fig 2)***

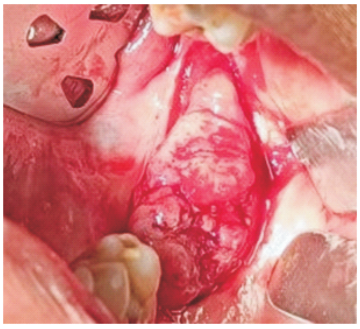

Fig 2: Intraoral view

It extended from the left first premolar region to 2.5 cm posteriorly anteroposteriorly and from the depth of left buccal vestibule up to the depth of lingual vestibule mediolaterally. It was painless and there was no bleeding on probing present. The swelling was covered by normal mucosa with no visual secondary pathological changes and the lesion was not associated with any regional cervical lymphadenopathy. Considering the relevant clinical findings, a provisional diagnosis of benign tumour of the left lower residual alveolar mucosa was made.

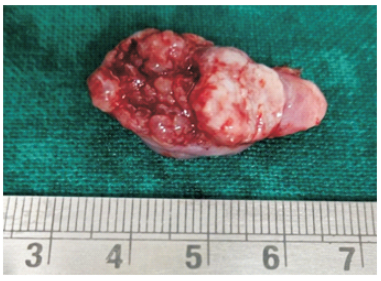

Fig 3: Excised Specimen

Incisional biopsy was done and the final diagnosis of recurrent Plexiform Ameloblastoma was made. The lesion was completely excised along with a healthy margin of 1.5 mm(Fig 3) and the specimen was sent for histopathological examination after fixation in 10% neutral buffered formalin.

The histopathological examination of the excised tumor revealed a Plexiform variant of Ameloblastoma, predominantly comprising of epithelium arranged as a tangled network of anastomosing strands. The cords or sheets of the epithelium are bounded by tall, columnar ameloblast like cells with reversed polarity surrounding more loosely arranged epithelial cells. The supporting stroma is also loosely arranged and vascular. These features were consistent with a diagnosis of plexiform ameloblastoma (Fig 4).

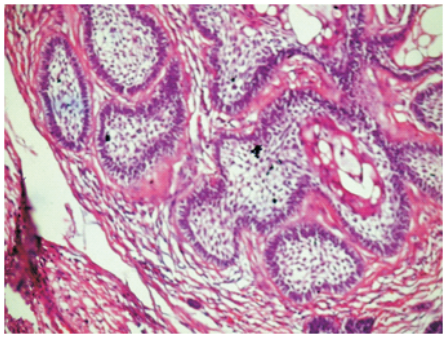

Fig 4: Microscopic view

After a follow-up period of nearly 23 months, the patient remains free of further recurrence.

Discussion:

Ameloblastoma is a benign epithelial odontogenic tumor but is often aggressive and destructive, with the capacity to attain a large size invading the adjacent vital structures. The first detailed description of this lesion was by Falkson in 1879 and the term ameloblastoma was first coined by Ivy and Churchill in 1960. It is the most common odontogenic tumor representing only about 1% of all the tumors and cysts of the jaws[5].

In the mandible, 70% of Ameloblastoma are located in the area of the molars or the ascending ramus,20% in the premolar region, and 10% in the anterior region[2]. About 10-15% of ameloblastomas are associated with a non-erupted or impacted tooth. In the present case, the plexiform ameloblastoma was found in the ascending ramus and molar region of the left mandible and it was not associated with a non-erupted tooth as the patient underwent Disarticulating Hemimandibulectomy followed by reconstruction using a Recon plate for the same reason 8years back.

The recurrence of ameloblastoma is of great concern. Reasons for recurrence include the locally invasive nature of the tumor, the clinical type of ameloblastoma (ie, solid vs unicystic), the histologic type, location (mandibular vs maxillary), and treatment modalities. In rare instances, the recurrence period for an ameloblastoma might be as long as 49 years, but 50% of ameloblastomas recur within[5] years after surgery[10]. In our study, recurrence was seen after 8 years.

Conservative treatment (enucleation or curettage) is the most unacceptable mode of treatment for multilocular lesions[11]. Unicystic ameloblastomas have much lower (20%) recurrence rates. About 75% to 100% of the solid tumors recur if treated conservatively, but only 18% or less recur when treated aggressively[12]. Of the different histologic variants, the follicular and granular types are associated with the highest recurrence rates[11]. The recurrence of plexiform variant is a rare case which is presented in this study.

In the present case, the recurrence was seen after an unexpected time frame of 8 years. Furthermore, on the basis of the microscopic features of the recurrent tumor, it was of the plexiform variant. Though recurrence some times occurs in bone grafts[13], it is unusual to find a recurrence in the soft tissues[14] as seen in our study.

The isolated soft tissue recurrence in this case was unusual and was probably due to tumour invasion into the adjacent soft tissue before the initial treatment which had not been clinically apparent. This case also demonstrated the ability of intraosseous ameloblastoma to grow into adjacent soft tissues. Adekeye EO [15] reported 38 cases of large ameloblastomas with tumour in contact with periosteum or adjacent soft tissue following cortical perforation which were managed with either marginal or segmental jaw resections..

We hypothesize that the surgical problem of excising the primary lesion, particularly from the mandibular ramus region, had probably left epithelial remnants in the soft tissues and caused the recurrence in this location and the tumor might have burrowed from its primary location through the mandibular ramus medially into the soft tissues extending anteriorly upto the premolar region. Nonetheless, it seems that the seeding of tumor cells in the native gingival soft tissues during surgery could be the main cause of recurrence

in the case investigated in the present study. The soft tissue recurrent tumors in our study was aggressive, and the pre-surgical radiograph of the primary lesion showed destruction of the cortical plate and possible infiltration into the adjacent soft tissues.

However, the high level of aggressiveness of this lesion was not noted at the time of initial surgery, and only radical bone resection was performed in our cases. We had also removed the overlying soft tissue as well during initial resection but still soft tissue recurrence was seen. Thus, from the present findings, it can be speculated that inadequate soft tissue removal or seeding during extraction of teeth 2 months before surgery or during surgery are most likely the causes of recurrence.

In conclusion, we propose to be extra-cautious during removal of ameloblastoma if patient has a history of tooth extraction in that area, as it may increase the chances of seeding of tumor cells. Hence, adequate excision of soft tissue margin near and around the extraction socket/cortical perforation should be performed to reduce the chances of recurrence.

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