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Unicystic ameloblastoma - out of the ordinary

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Abstract

The ameloblastoma is the prototype of a benign neoplasm—that is, it will exhibit cell replication and growth throughout its existence, but it will not metastasize[1]. Ameloblastomas are the true neoplasm of the enamel organ which does not differentiate to the level of enamel formation. Ameloblastomas are a varied entity which can be further classified based on the nature of invasion and the surgical approaches recommended for each type as ameloblastoma in situ which has mural and intraluminal variants, second is micro invasive ameloblastoma which has intramural and transmural variants, third and the last type is invasive ameloblastoma which has invasive and invasive type arising from the lining of a cyst. Here we present the case of a 59-year-old patient diagnosed with ameloblastoma in situ treated with enucleation and chemical cauterization with carnoys solution.

Keywords: Ameloblastoma, Cystic, Cavity

Introduction

The term "adamantinoma" was coined by Malassez in 1885. Since the term implied the formation of hard tissue, and no such material was present in this lesion, it was replaced with the term "ameloblastoma" as suggested by Churchill in 1934[2]. Unicystic ameloblastoma, which is also known as the ameloblastoma in situ, has an asymptomatic growth and progress which in most cases is reported to give a painless pattern of development. It is a lesion that if left untreated can increase in size and further lead to esthetic

disfigurement combined with functional discrepancies which makes the diagnosis and treatment to become inevitable. This is pathologically a rare occurring entity with 2-3% of occurrence, with its peak occurrence in 20-30 years[1], mandibular molar effected in 75% and maxillary molar effected in 25% of the patients, which makes the case presented in this article a rare one with its out of ordinary age relevance and site of occurrence. A high percentage of these lesions are associated with an impacted tooth and the most commonly cited provisional diagnosis is dentigerous cyst[2].

Case report

A 59-year-old female patient who is a daily wage worker presented with a chief compliant of intraoral swelling over the upper right jaw region for 1 year. Patient had undergone extraction of 15 and 16 in a private clinic because of the swelling 6 months back and no change in the size of swelling was noted even after the extraction procedure. Extra orally, no symptoms of pain or paresthesia or heaviness of head noted. Intraorally, inspectory findings noted are swelling in the vestibular region of 15 and 16, which is approximately 5x5 cm in size (fig 1). Palpatory findings revealed it to be non-tender, non-fluctuant, sessile swelling. Panoramic examination showed the swelling to be of a Unicystic type extending from the apical part of 14 to the mesial part of 18 with missing 15, 16 and 17(fig 2). Radiographically, the lesion was seen to be extending to the maxillary sinus. Computed tomography was also taken to confirm the location and extension of the lesion. (fig 3)

A biopsy under local anesthesia was carried out after blood investigations taking into account of patients age into consideration (Haemoglobin=11.8 gram %; RBC= 4.2 million mm^3; WBC= 4550 mm3). Aspiration biopsy revealed a yellowish coloured fluid with crystals (fig 4). The lining of the cyst was exposed, a part of it was resected and sent for further histopathological investigation and the result obtained was Unicystic ameloblastoma.

Surgical enucleation was carried out under general anesthesia after obtaining surgical fitness of the patient from the physician. Written consent from the patient was taken before the operation. Cystic lining was identified and removed out in toto (fig 5 and fig 6); Sinus was seen to be pushed upwards with no perforations inside. Chemical cauterization was performed with freshly prepared modified carnoys solution (fig 7). Thorough irrigation was done with around 150ml of normal saline solution and then irrigation with metrogyl solution was carried out. The cystic cavity was filled with the PRF and then wound sutured back carefully (fig 8).

Postoperative pressure dressing was kept for 48 hours and intravenous antibiotics and analgesic for next 3days followed by 2 days of oral antibiotics and analgesics. Suture removal was carried out on the 10th post-operative day.

Discussion

Ameloblastomas are entities which have their origin from various sources:

- Cell rests of the dental organ, either remnants of the dental lamina (cell rests of Serres) or remnants of the Hertwig's epithelial root sheath (epithelial rests of Malassez).
- Epithelium of odontogenic cysts, particularly dentigerous cyst.
- Disturbances of the developing enamel organ.
- Basal cells of the surface epithelium of the jaws[2].

When it comes to specific details about Unicystic ameloblastoma, it was first described by Robinson and Martinez in 1977[10].

Ackerman's classification of Unicystic ameloblastoma into three histologic groups is as follows:

- Luminal UA (tumor confined to the luminal surface of the cyst);
- Intraluminal/plexiform UA (nodular proliferation into lumen without infiltration of tumor cells into connective tissue wall)
- Mural UA (invasive islands of Ameloblastomas epithelium in the connective tissue wall not involving the entire epithelium[4].

According to this classification, our lesion is the mural type (fig 9).

There is another grouping by Philipsen and Reichart which described the forms of Unicystic Ameloblastoma as follows:

Subgroup 1.: Luminal UA;

Subgroup 1.2.: Luminal and intraluminal;

Subgroup 1.2.3.: Luminal, intraluminal and intramural; and

Subgroup 1.3.: Luminal and intramural[4].

For Unicystic ameloblastoma, the histopathological examinations are very much valid because of the variations they exhibit. The mural variant shows hyperchromatism of basal cell nuclei, palisading and polarization of the basal cell nuclei and cytoplasmic vacuolation of basal and basilar cells.

Intercellular spacing of the epithelium is also present, and basal budding may occur[1].Intraluminal variant shows intraluminal proliferations and microinvasive Ameloblastomas contain islands of ameloblastic epithelium in the cyst wall. Usually the root resorpotion, if present in ameloblastoma in situ cases, will have a smooth and regular resorption pattern. Despite displacement of the inferior alveolar canal, nerve sensation is not altered by the tumor because of its inability to undergo true neural invasion[1].

When it comes to the treatment of Unicystic ameloblastoma, we have a lot of treatment options. Enucleation marsupialization or even resection based on the site, severity and functional parameters. Enucleation is the most commonly applied treatment plan for Unicystic ameloblastoma but according to MARX et al ameloblastoma can extend from 2.3 to 8 mm beyond the radiographic margin of the tumour, thus, by enucleation alone, the ameloblastic cells will be left behind despite the tumour being enucleated whole[10.] When enucleation is combined with other techniques chemical cauterization or cryotherapies, the result is better with less recurrence. Carnoy's solution, a powerful tissue fixative that can penetrate bone to 1.54 mm, was first described by culter& Zollinger in 1933 as a treatment for cystic lesions[10].Marsupilization is the treatment modality when recurrence is not a concern and patient is Severly ill, it will give shrinkage of the cyst. Resection is the treatment plan with least chance of recurrence but patients age, Esthetic features and the quality of life should be taken into account.

Differential diagnosis

Usual appearance of a Unicystic ameloblastoma seen in orthopantomogram is a unilocular radiolucency which may or may not be associated with impacted teeth, which gives a diagnosis as dentigerous cyst. Odontogenic keratocyte can also be a differential diagnosis which can be co-related with clinical features. Adenomatoid odontogenic tumor can also be a variation in diagnosis if the age is below 20 years and the site is in anterior maxillary jaw. Calcifying odontogenic cyst usually is associated with calcification in the radiographs but if it's not associated with any calcified structures, then it can also be included.

Recurrence

Enucleation alone yielded the highest recurrence rate among all treatments (30.5%)[10]. Resection for Unicystic ameloblastomaresults in the lowest recurrence rate (3.6%)[10]. Marsupialization together with other treatments resulted in an 18% recurrence rate[10]. Enucleation followed by the application of Carnoy's solution resulted in a recurrence rate of 16%, which was the best except for resection[10].

Conclusion

When a Unicystic case is planned for treatment, the histological variant identification becomes a very important factor for proper planning of the treatment. The mural type has the highest recurrence rate because of its nature to penetrate the epithelium. Li et al reported a higher recurrence rate of 35.7% for the mural type and a lower recurrence rate of 6.7% for the luminal and intraluminal types[6]. The modality of planning should be multifactorial depending upon the site, size, Esthetic balance and functional rehabilitation. After the operative procedures it's very important to give a proper follow-up for the patient to analyse any changes of recurrence. More than 50% of cases recurred within 5 years after the operation.

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Legend Figures

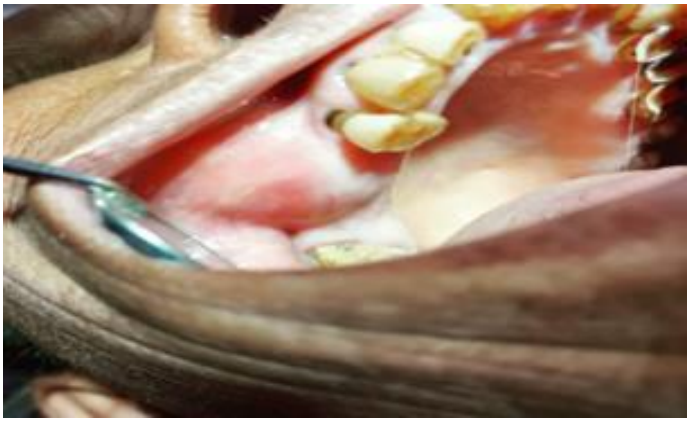

Figure 1: Pre-operative photograph

Figure 2: Pre-Operative orthopantamogram

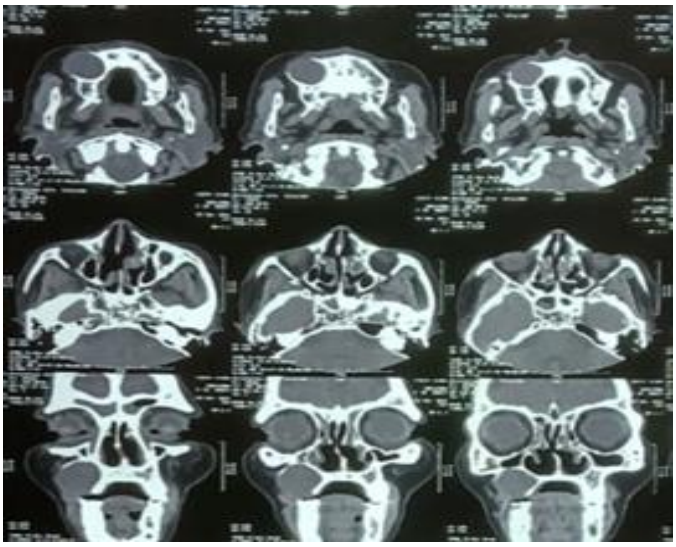

Figure 3: CT view of the lesion

Figure 4: Aspirated contents of the biopsy procedure

Figure 5: lining of the Unicystic Ameloblastoma

Figure 6: Cystic cavity after the removal of the lining

Figure 7: Cystic cavity treated with Carnoys Solution

Figure 8: Wound closure

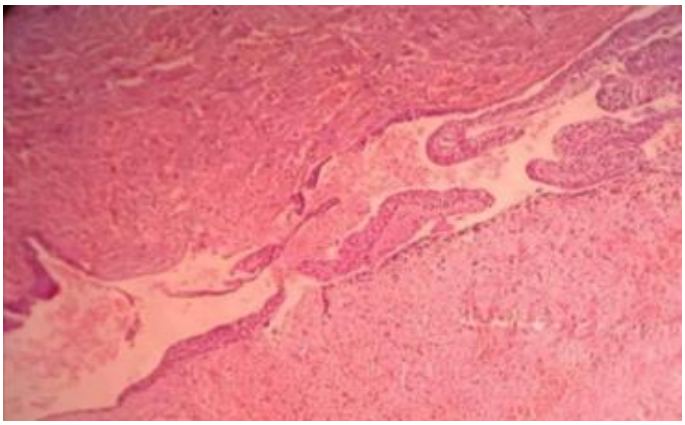

Figure 9: Histopathologic photographs of the excised specimen