Management of Plexiform Ameloblastoma in a 12 year old female: A Case Report

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Abstract

Ameloblastoma is a true neoplasm of odontogenic origin. Ameloblastoma is responsible for 1% of all the oral and maxillomandibular cysts and tumors. It is odontogenic in origin and benign in nature but it has a high percentage of local recurrence rate and possible malignant development when treated inadequately. We report a case of plexiform ameloblastoma presenting in a 11-year-old female. The aim of this article is to evaluate the clinical result of the patient reported to us with mandibular ameloblastoma using conservative management. The tumour was conservatively managed by enucleation and chemical cauterization. Histological analysis demonstrated a plexiform ameloblastoma. The patient remains well without disease after 3 years of postoperative follow-up and still being followed up.

Keywords: Ameloblastoma, maxillomandibular cysts and tumors, plexiform ameloblastoma, enucleation, chemical cauterization.

Introduction

Ameloblastoma is a benign epithelial odontogenic tumor but is often aggressive and destructive, with the capacity to attain great size, erode bone and invade adjacent structures [1]. The World Health Organization (1991) defined ameloblastoma as a benign but locally aggressive tumour with a high tendency to recur, consisting of proliferating odontogenic epithelium lying in a fibrous stroma [2].

It represents about 1% of all oral ectodermal tumors and 9% of odontogenic tumors [3]. Most ameloblastomas develop in the molar-ramus region of the mandible with 70% of these arising in the molar-ramus area and they are occasionally associated with unerupted third molar teeth [4]. Ameloblastoma appears most commonly in the third to fifth decades but the lesion can be found in any age group including children [3].

Case Report

A 12-year-old girl was referred to the department of oral and maxillofacial surgery, with a diffuse swelling in the right side of mandible [Figure 1]. The swelling was painful and had been slowly increasing in size for 6 months. Extra oral examination revealed the swelling measuring about 4 cm x 4 cm in size on the right mandible extending antero-posteriorly to about 4 cm from the symphysis region to 1 cm in front of the ear lobule, and supero-inferiorly it was 2 cm from the cheek prominence to 1 cm below the lower border of the mandible. The swelling was firm in consistency, tender on palpation and a mild rise in temperature was evident.

Intraorally, the swelling extended from the distal surface of right first molar to the retromolar region, obliterating the buccal vestibule [Figure 2]. The orthopantomograph revealed a well-defined unilocular radiolucency extending from distal to the first molar region towards the ramus of involving the body ramus and coronoid process of the mandible on the involved side [Figure 3]. It was found that second molar was impacted and radiolucency was present around its crown portion. Displacement of the roots of first molar was evident. Paranasal sinus radiography revealed a well-defined radiolucency involving the ramus of mandible with its bucco-lingual extensions [Figure 4].

Fine needle aspiration cytology was performed which did not give any conclusive evidence. Therefore, an extensive enucleation of the said lesion along with sub-periosteal dissection was planned under general anesthesia. A Modified Wards incision was placed using a No.15 B.P. blade, extending anteriorly up to the region of canine. After reflecting the mucoperiosteal flap, the expanded cortical plate was identified and separated from the mucoperiosteum. Subperiosteal dissection was carried out beginning from the sound bone near the canine region to posteriorly over the ramus of mandible [Figure 5]. The mental nerve was identified and preserved. The cystic lining was separated from the inferior border of the mandible taking care not to injure the inferior alveolar nerve. The impacted tooth bud was delivered out with
its cystic lining. The retracted cystic mass was then sent for histopathological analysis. The histopathological processing of the tumor revealed a plexiform ameloblastoma predominantly composed of epithelium arranged as a tangled network of anastomosing strands enclosing cysts of various sizes [Figure 6]. Based on these findings a diagnosis of unicystic plexiform ameloblastoma was made.

Discussions and Conclusion

Ameloblastoma is a true neoplasm of odontogenic epithelium. It is uncommon in children, in a review of 1,036 ameloblastomas of jaw, the average patient age is 38.9 years, with only 2.2% (19 of 858) were under 10 years and 8.7% (75 of 858) were between 10 and 19 years [5]. Ameloblastomas are slow growing and locally invasive tumors, occurring in three different clinicoradiographic situations namely, Conventional solid/multicystic, unicystic and peripheral [6].

Typical ameloblastoma starts insidiously as a central bony lesion which is slowly destructive; however tends to expand the bone instead of punching a hole through it. The tumor is rarely painful, unless infected and usually does not cause signs and symptoms of nerve involvement, even when large. Ackerman et al, in their study of unicystic ameloblastomas, defined three subgroups. Group I (42%) consisted of a unicocular cyst with a nondescript but variable epithelial lining. Inactive odontogenic cell rests might be present in the fibrous wall, but there was no infiltration by neoplastic epithelium. Group II lesions (9%) featured intraluminal plexiform proliferation but no infiltration of the cyst wall. In Group III lesions (49%), plexiform or follicular-type ameloblastoma, sometimes in continuity with the cyst lining, infiltrate the wall [7].

Solid ameloblastoma is the most common form of the lesion (86%). It has a tendency to be more aggressive than the other types and has a higher incidence of recurrence [8]. Unicystic ameloblastoma has a large cystic cavity with luminal, intraluminal or mural proliferation of ameloblastic cells. It is a less aggressive variant and it has a low rate of recurrence, although lesions showing mural invasion are an exception and should be treated more aggressively. Peripheral ameloblastoma exists in soft tissue. Treatment of mandibular ameloblastoma continues to be controversial. Prior to choosing a treatment for ameloblastomas, the clinicoradiologic variant (solid, multicystic, unicystic, peripheral), anatomic location, clinical behavior and size of the tumor, and age of the patient should be assessed. Besides surgery, treatment may also include cryo-radio and chemotherapy. When treated inadequately, malignant development is a possibility.

The tumor found in our patient was an ameloblastoma of the plexiform type. The term “plexiform” refers to the appearance of anastomosing islands of odontogenic epithelium in contrast to a follicular pattern. Unicystic ameloblastomas have been shown to have less recurrence (15-48%). Histologically, it presents cystic characteristics delimited by a layer of ameloblastic epithelium. There are three types of unicystic ameloblastomas: intraluminal, plexiform (where enucleation is considered the treatment indicated) and mural, that requires marginal resection because of aggressive behaviour and higher recurrence rate [9].

The histological patterns have no prognostic validity, except for unicystic subtypes. The unicystic ameloblastoma usually presents between 16 and 20 years of age, and the multicystic ameloblastoma after 30 years of age. Generally, the unicystic ameloblastoma presents an unicocular image associated to third molars [10]. Considering the age of the patient, and the various studies a conservative approach was used in this patient, rather than a radical approach. More such cases should provide us an insight to the biologic behavior and clinical course of such tumors, which may help us in effective treatment plan.

References

Illustrations

Illustration 1

Figure 1

Illustration 2

Figure 2
Illustration 3

Figure 3

Illustration 4

Figure 4
Illustration 5

Figure 5

Illustration 6

Figure 6
Reviews

Review 1

**Review Title:** Management of Plexiform Ameloblastoma in a 12 year old female: A Case Report

Posted by Dr. Fayyaz Ahmed on 10 Dec 2011 11:16:28 AM GMT

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**Rating:** 7

**Comment:**
The article is written quite well, but minor grammatical corrections to be made. The author has to include some more references in discussion as this topic is vague and there still exists a dilemma in management.

**Competing interests:** non

**Invited by the author to make a review on this article?** : Yes

**Experience and credentials in the specific area of science:**
am assistant professor in dept of oral and maxillofacial surgery where i teach the subject to the under and post graduates

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