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REVIEW ARTICLE

Bilocular unicystic ameloblastoma of the mandible in a 9 yr old child – A diagnostic and management dilemma

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KEYWORDS

Unicystic ameloblastoma;
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Enucleation

Abstract Unicystic ameloblastoma is a less encountered variant of the ameloblastoma that usually presented as unicystic lesions of jaw occurring in 3rd and 4th decades of life. It shows a typical ameloblastomatous epithelium lining the cyst cavity, with or without variable tumor proliferations. The case presented here is of a 9 yr old boy who was referred to our center for the management of a large diffuse swelling on the right side of the face. Clinical and radiologic evaluation showed two interconnected cystic lesions in the right body and the symphyseal regions of the mandible associated with impacted canines bilaterally. The initial histopathology of both cystic spaces showed the lesion to be dentigerous cysts and the results were reconfirmed in two other centers. A complete surgical enucleation of this bilocular cyst was done sparing the impacted teeth. The histopathologic examination of the post-operative specimen showed features of Unicystic Ameloblastoma. The patient was followed up on a regular basis for more than 3 years. There is no signs of recurrence and his latest radiographic examinations shows good bone formation. The impacted teeth are erupting into position. This case reports the difficulty in clinical diagnosis and the peculiar bilocular presentation of unicystic ameloblastoma which was conservatively managed by surgical enucleation of the complete lesion, sparing the dentition.

Contents

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1. Introduction

Ameloblastomas are characterized as slow growing, expansive odontogenic tumours with a radiolucent, uni or multilocular (soap bubble) radiographic appearance, that tend to infiltrate surrounding tissues (Amzerin et al., 2011). It is also called as multilocular cyst of jaw. However, if the tumour presents clinically as a unilocular cystic lesion it is classified as a unicytic ameloblastoma (UA). Robinson and Martinez in 1977 were the first to describe unicystic ameloblastoma defining it as a cystic cavity lined by ameloblastic epithelium with three different variants: luminal, intraluminal and mural (Hollows et al., 2001; Mahajan et al., 2014). Even though this terminology was adopted by the WHO in 1992, the name "cystogenic ameloblastoma" is also recognised by WHO (Bajpai et al., 2013).

Radiographically, unicystic amaeloblastomas may resemble a dentigerous cyst or a kerato cystic odontogenic tumour (KCOT) and therefore pose a difficulty in clinical diagnosis (Mohanty et al., 2013). The multilocular appearance in unicystic ameloblastomas were also reported. (Bajpai et al., 2013) However a bilocular appearance of UA in two anatomic regions of mandible is not previously reported. Anatomically amaeloblastomas are commonly seen in the molar and the ramus region of the mandible, and are noticed in the 3rd and 4th decades of life, with equal sex predilection (Chaudhary et al., 2011).

The treatment of large multiple cystic lesions in children are always a challenge to the surgeon in terms of diagnosis and treatment plan to decide whether an aggressive resection, complete enucleation or conservative marsupialisation is the most appropriate option. (Arora et al., 2013) UA of mandible is a very rare entity in under 10-year paediatric population (Philipsen and Reichart, 1988). Its clinical and radiological appearance along with unerupted permanent teeth in children gives a picture suggestive of dentigerous cyst. A histopathological study by initial incision biopsy guides clinician to confirm the diagnosis and the treatment plan. However larger lesions often exhibit ameloblastomatous epithelial proliferation of the cyst wall which is not always detected in initial incision biopsy. Here we are reporting a case of two interconnected cystic lesions of mandible in a 9-year-old boy which were diagnosed to be dentigerous cysts during their incision biopsies. The detailed histopathologic study after surgical enucleation of the both cystic spaces showed the lesion showing features of Unicystic ameloblastoma. The diagnostic dilemma and the effective management of such lesions in children are described and the histopathological variations are discussed

2. Case report

A 9-year-old Saudi boy was referred to the maxillofacial clinic at King Fahd Military Medical Complex, for the management of a large swelling on the right side of the face. The boy had noticed the swelling approximately one month back, following a fall while playing football. The swelling was initially diagnosed as a dentoalveolar abscess and therefore an incision and drainage and extraction of the decayed lower right first molar was done at the referring hospital. He was well nourished for his age, medically fit and all his blood investigations done in the previous centre were within normal limits.

On arrival at our centre, the boy presented with a diffuse large swelling extra orally on the right mandibular region, associated with tenderness on palpation (Fig. 1). On intra oral examination his lower right molar extraction socket was healing well, with no signs of any infection. There was bony expansion of the labial cortex from the lower right 2nd molar region to the lower left canine region. There was no sign of any facial nerve or inferior alveolar nerve weakness. An orthopantomogram showed two large cystic lesions one in the right body of the mandible and the other at the symphysis of the mandible. The lower left 2nd molar and the lower bilateral canines were unerupted. (Fig. 11). CT scan showed two cystic lesions involving one in the entire right body of the mandible and the second at the symphysis of the mandible connected by a canal like structure around intact bone in the right canine region (Figs. 2 and 3) Two incisional biopsies were done to get a confirmation of the both lesions appeared in the x-rays. Two separate specimens were taken, one from the posterior lesion and the other from the symphysial lesion. The initial

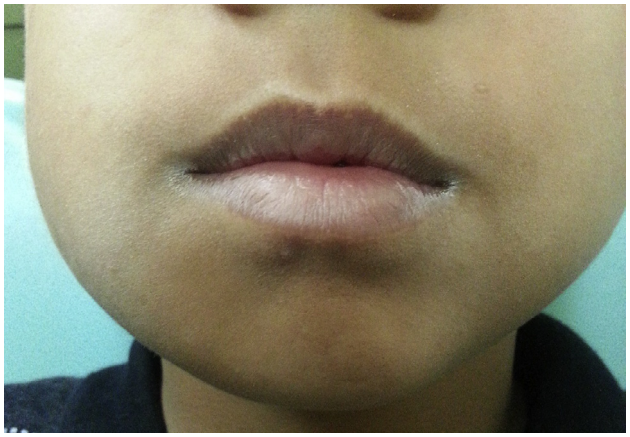

Fig. 1 Pre-operative clinical appearance.

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