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Case Report

Ameloblastoma of the Mandible in a 13-Year-Old Female—Case Report

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ABSTRACT:

The ameloblastoma according to the classification of odontogenic tumors by WHO in 2005, is classified as a benign neoplasm of odontogenic epithelial origin. One to three percent of tumors and cysts of the jaws are comprised of ameloblastomas. The tumor is locally aggressive, but often asymptomatic, showing a slow growth which is manifested as a facial swelling or radiographic incidental finding. It is a benign epithelial tumor that has aggressive, destructive and unlimited growth potential, having the capacity for recurrence and malignant transformation. Regarding the symptoms and clinical signs, the presentation of ameloblastoma is poor. In children and young people, ameloblastoma can be difficult to diagnose, because it mimics other benign lesions. Its diagnosis requires a combination of imaging data, histopathological analysis and molecular tests. The methods of treatment consist of radical surgery (segmental resection) and conservative treatments (enucleation with bone curettage). The particularity of the presented case is that it was represented by the late request as getting the treatment for swelling but it was still persisting. Secondly, the continued growth and facial bone physiology (higher percentage of cancellous bone, increased bone turn over and high periosteal reactivity), as well the presence of unerupted teeth.

Keywords: ameloblastoma, odontogenic tumors, multiloculated cystic lesion

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INTRODUCTION

The ameloblastoma according to the classification of odontogenic tumors by WHO in 2005, is classified as a benign neoplasm of odontogenic epithelial origin (Barnes et al., 2005). One to three percent of tumors and cysts of the jaws are comprised of ameloblastomas (Small & Waldron, 1995; Reichart et al., 1995). Ameloblastoma is the most common odontogenic tumor (OT) in Africa (Arotiba et al., 1997; Ladeinde et al., 2005) and Asia (Wu & Chan, 1985) but is the second most common in South and North America (Regezi et al., 1978; Ochsenius et al., 2002). Ameloblastoma can theoretically arise from remnants of the dental lamina, enamel organ of developing tooth, the epithelial lining of odontogenic cyst or basal cells of the oral mucosa (Crawley & Levin, 1978; Leider et al., 1985). It occurs in almost all age groups, but mainly diagnosed in the third or fourth decade of life. Most cases (66%) affect the posterior mandible and ramus (Neville et al., 2008). Ameloblastomas are usually asymptomatic and present as a slow growing facial swelling or as an incidental radiographic finding. Despite being a benign neoplasm, it is locally destructive and has a high rate ofrecurrence if not completely removed (Hong et al., 2007). The three clinical and radiographic presentations which have different prognostic and therapeutic considerations can include: 1) solid/ multicystic (86% of cases); 2) unicystic (13% of cases); 3) peripheral (1% of cases) (Neville et al.). Its classic radiographic presentation is that of a multilocular radiolucency. The expansion of the buccal and lingual cortices of bone, with the possibility of bone perforation and soft tissue extension is frequently observed. The resorption of roots of adjacent teeth is common and is often associated with an unerupted tooth. Most frequently, it is the mandibular third molar area which is involved (Dunfee et al., 2006). However, the solid/multicystic ameloblastoma may appear radiographically as a

unilocular lesion resembling other cystic lesion (Hong et al.) The clinico-pathologic characteristics are of a benign lesion with a slow growth pattern, but locally invasive. The clinical behavior can be considered between a benign and malignant lesion, and the high rate of recurrence is an important factor when determining the management of the lesion (Chapelle et al., 2004).

Therefore, the choice of treatment should be assessed based on the lesion's clinical type (solid/multicystic, unicystic, peripheral), the location and size of tumor and patient's age. The spectrum of treatment described in the literature range from simple bone curettage to segmental resection, but there are few criteria for treatment based on retrospective studies published. In this report we present the unusual case of a solid/multicystic ameloblastoma in the mandible of a 13-year-old girl.

CASE REPORT

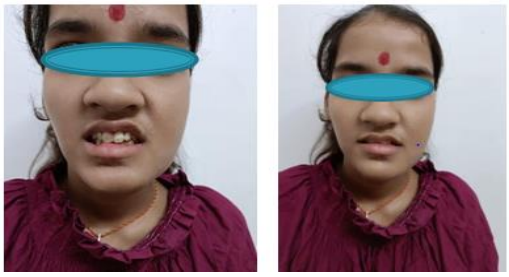


Fig. 1. a-b. Extraoral photographs showed facial swelling over the body of the left mandible.

The patient is a 13-year-old female, without history of medical conditions. She was consulted to the Oral and Maxillofacial Surgery department of tulsi dharnia hospital, Bikaner, Rajasthan due to a painless facial swelling in the left perimandibular area for the last one years. The particularity of this case is represented by the late request for medical consultation.

Extraoral : clinical examination showed a mild facial swelling over the body of the left mandible, which was firm to palpation with a normal overlying skin (Fig. 1).

Intraorally, the examination was remarkable for a buccal and lingual expansion of the mandibular left body, tender to palpation and covered with normal, healthy mucosa (Fig. 2). There were neither palpable neck masses nor lymphadenopathy and all cranial nerves were intact. The remaining physical exam was within normal limits. The patient had no relevant medical history and was taking no medication.

Computed tomographic, CT, images, usually show an expansile, radiolucent, multiloculated cystic lesion, with a characteristic ''soap bubble-like'' appearance. The computed tomography (CT) requested, revealed an extensive multilocular and radiolucent lesion with diffuse margins, localized to the left mandibular body extending from the second premolar to the first molar (3.6 cm antero-posterior and 2.3 cm width) . Reabsorption of the roots of adjacent teeth and expansion of the buccal and lingual cortical plates with evident perforation in some areas were also noted (fig .2)

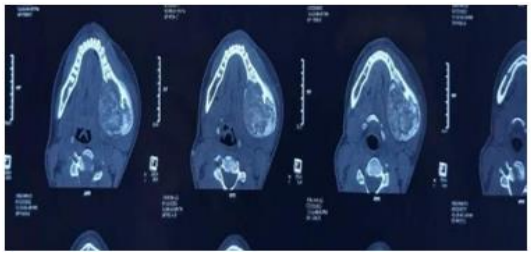


Fig.1

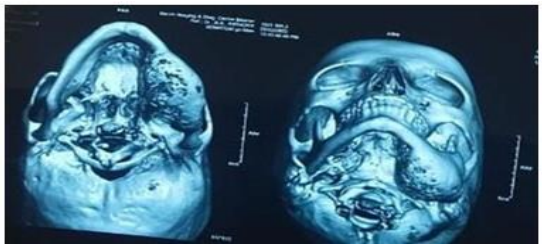
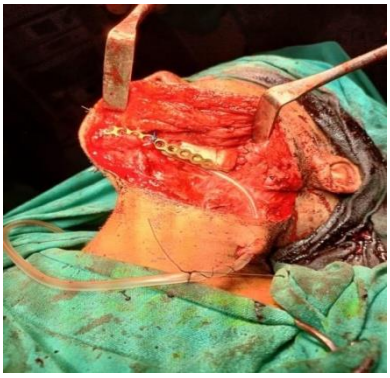


Fig.2

The clinical and imagistic findings could have been suggestive of ameloblastoma. The main differential diagnosis that was considered was an odontogenic keratocyst. In this case, the risk of pathological fracture of the mandible is increased. It was decided that the patient should undergo emergency surgery, which is why no preoperative biopsy was performed. The lesion was surgically enucleated and curettage.

The harvested specimens sent to pathology services and processed with the usual histological technique. The histopathologic examination demonstrated continuing islands of odontogenic epithelium set in a fibrous stroma. The epithelium consisted of basal cells resembling the enamel organ, showing cytoplasmic vacuolization and reverse polarization of the nuclei that resulted in solid/multicystic ameloblastoma. The patient underwent treatment with a mandibular resection with safety margins of 1 cm, through a submandibular approach along with placement of a 2.4mm mandibular reconstruction plate (Fig. 3).

Submandibular approach to mandible



Mandibular resection and reconstruction plate placement
Fig .3

DISCUSSION

Ameloblastoma is a benign epithelial tumor that has an aggressive, destructive, unlimited growth potential, having the capacity for recurrence and malignant transformation.

The diagnosis requires a combination of imaging data and histopathological analysis to be confirmed. The methods of treatment consisted of radical surgery (segmental resection) and conservative treatments (enucleation with bone curettage). The particularity of the presented case is represented by the late request for medical consultation.

Ameloblastoma is a benign epithelial tumor that constitutes about 14% of all jaw tumors and cysts. It has aggressive, destructive and unlimited growth potential, having the capacity for recurrence, malignant transformation and metastasis (in approximately 1% of cases). There is no differentiation according to sex. The global incidence of ameloblastoma is 0.5 cases/million people, with 10–15% of cases occurring in the pediatric population reaching up to 25% ~80% of cases occur in the mandible and ~20% cases occur in the maxilla . Very rarely is it reported in other head and neck sites like the sinonasal tract, middle ear, temporal bone and infratemporal fossa.The maximum incidence depending on age varies as follows: conventional type ameloblastoma, between 40 and 50 years; unicystic type ameloblastoma, between 20 and 30 years; and extraosal/peripheral type ameloblastoma, between 50 and 70 years. Regarding the symptoms and clinical signs, the presentation of ameloblastoma is poor.

In some cases, a radiological change is occasionally detected on radiographs taken for other reasons. Painless swelling, with slow regional bone growth, is the most common presenting symptom of ameloblastoma. Invasion of soft tissues, mobility of adjacent teeth and dental malocclusion are other clinical signs. Pain is an unusual symptom that can occur as a result of hemorrhage inside or adjacent to the tumor, or as a result of the invasion of some nerve structures.

In children and young people, ameloblastoma can be difficult to diagnose, because it mimics other benign lesions Ameloblastoma treatment in children would be complicated by 3 factors (Ord et al.):

1. The continued growth and facial bone physiology (higher percentage of cancellous bone, increased bone turnover and high periosteal reactivity), as well the presence of unerupted teeth.
2. Difficult initial diagnosis.
3. Predominance of AB unicystic type.

The diagnosis of ameloblastoma in children is difficult because most of the lesions Radiographically resemble dentigerous cyst. Studies associate ameloblastoma with an unerupted tooth in the range of 70% to 83%, and in our case is associated with a second premolar but is most frequently associated with the mandibular third molar.

The basis of treatment in adults is surgery, resection with safety margins of 1-1.5 cm is recommended due to the high rate of recurrence of the solid/multicystic ameloblastoma. The recurrence rate after resection is close to 5% compared with the 90-100% of the curettage and enucleation.

Ord et al. states that the solid/ multicystic ameloblastoma or recurrent lesions in children should be treated with mandibular resection in the same way adults are treated. Their cases are treated with mandibular resection with a safety margin of 1 cm of cancellous bone and soft tissue resection if there was cortical perforation. Our reported case was treated in this way with reconstruction plate placement. Monitoring is essential in these patients because most recurrences occur within the first 5 years.

The prognosis for ameloblastoma varies depending on age, type, location and size of the formation, in direct relation to the degree of bone involvement, damage to adjacent structures and type of surgical intervention (radical or conservative).

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