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Mandibular Ameloblastoma in a 10-year-old Child: Case Report and Review of the Literature

Ameloblastoma Mandibular en Niño de 10 Años: Reporte de un Caso y Revisión de la Literatura

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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. On clinical examination, the tumor can cause symptoms such as pain, ulceration, tooth mobility, root resorption and malocclusion. Ameloblastomas have a high rate of recurrence if not completely removed. It occurs in almost all age groups, but is mainly diagnosed in the third or fourth decade of life. The tumor is very rare in children. We present an unusual case of a solid/multicystic ameloblastoma of the mandible in a 10-year-old girl. In addition, a brief review of the literature on reported cases of this pathology in children is also presented.

KEY WORDS: solid ameloblastoma, multicystic ameloblastoma, odontogenic tumor, children.

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 of recurrence 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*.).

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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 10-year-old girl. In addition, a brief review of the literature on reported cases of this pathology in children is also presented.

CASE REPORT

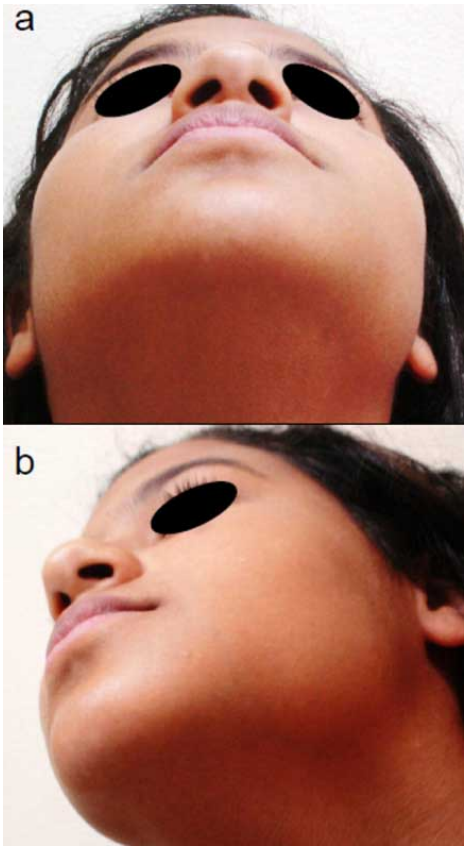

Fig. 1. a-b. Extraoral photographs showed mild facial swelling over the body of the left mandible.

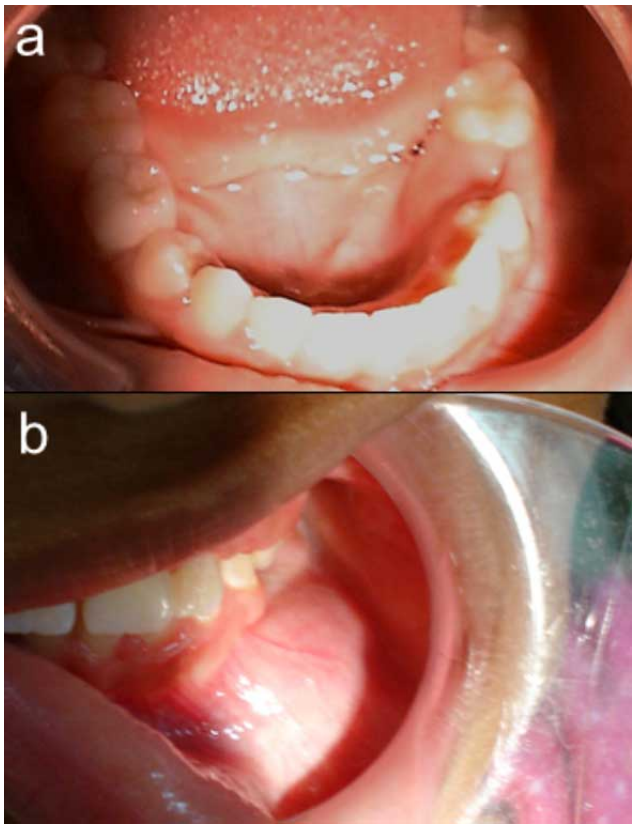

Fig. 2. a-b. Intraoral photographs showed remarkable buccal and lingual expansion of left mandibular body.

The patient is a 10-year-old Hispanic female, without history of medical conditions. She was consulted to the Oral and Maxillofacial Surgery Clinic at the University Pediatric Hospital (UPH) at the Medical Center in San Juan, Puerto Rico, due to a painless facial swelling in the left perimandibular area with three months of evolution. 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 exam 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.

The panorex and maxillofacial computed tomography (CT) requested revealed an extensive unilocular and radiolucent lesion with diffuse margins, localized to the left mandibular body extending from the canine to the first molar (3.6 cm antero-posterior and 2.3 cm width) and including the second premolar inside the lesion. Reabsorbtion of

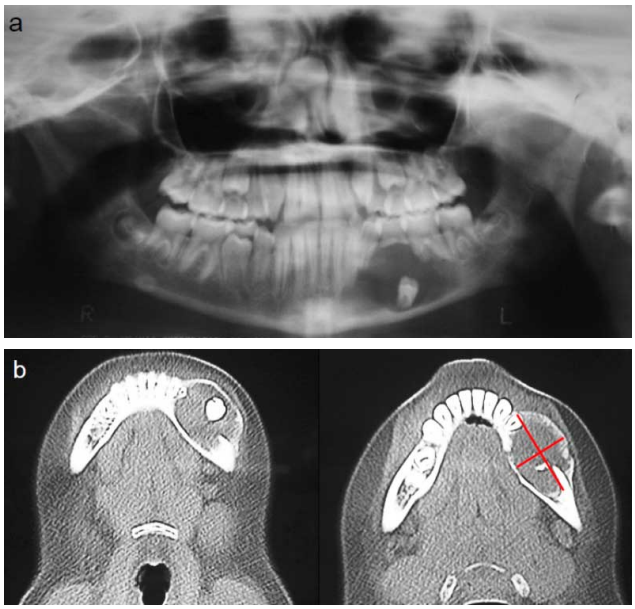

Fig. 3. a- Panorex showed extensive unilocular, radiolucent lesion of left mandibular body and including the second premolar inside the lesion, b- CT axial views of bone window, the dimensions of the lesion were: 3.6 cm antero-posterior and 2.3 cm width.

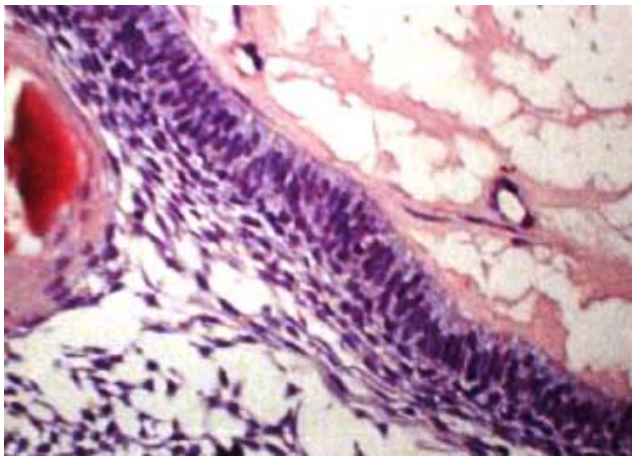

Fig. 4. Histopathology, hematoxylin-eosin stain, original magnification 100x.

the roots of adjacent teeth and expansion of the buccal and lingual cortical plates with evident perforation in some areas were also noted (Fig. 3).

The primary differential diagnoses contemplated for this lesion included an odontogenic cyst vs. an odontogenic tumor, however, a vascular lesion has to be always considered. The lesion was first aspirated prior to the extraction of the lower left deciduous first molar, through which an incisional biopsy was performed. Diagnostic hypothesis included:

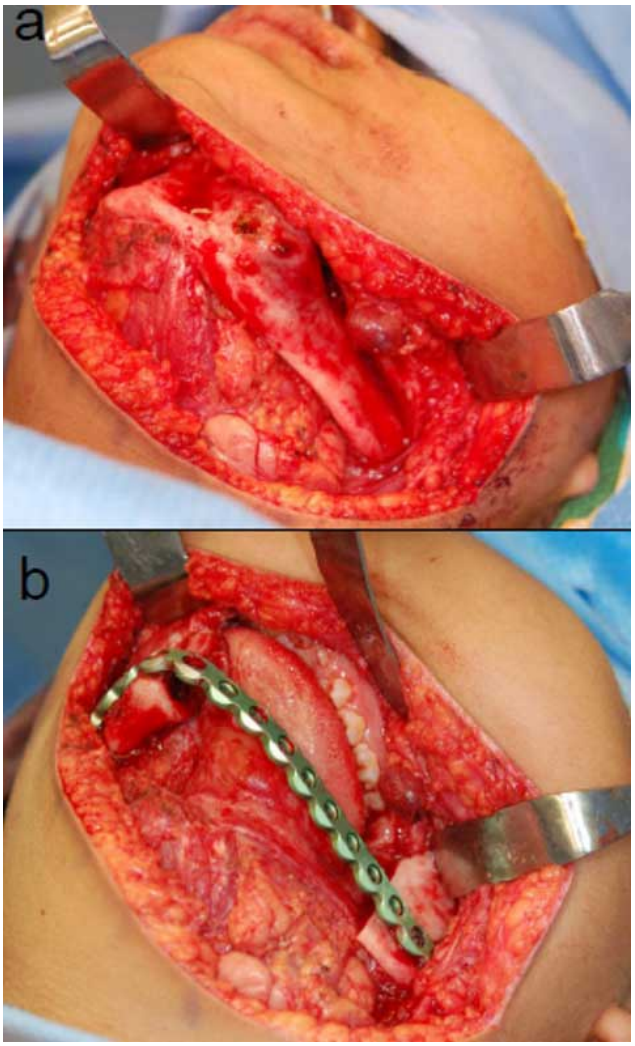

Fig. 5. a. Submandibular approach to mandible. b. Mandibular resection and reconstruction plate placement.

dentigerous cyst, primordial cyst, adenomatoid odontogenic tumor, keratocystic odontogenic tumor and unicystic ameloblastoma.

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 (Fig. 4). The patient underwent treatment with a mandibular resection with safety margins of 1 cm, through a submandibular approach along with placement of a 2.4 mm mandibular reconstruction plate (KLS Martin LP, Jacksonville, Florida) (Fig. 5). Specimen X-Ray was obtained to check the bony margins (Fig. 6).

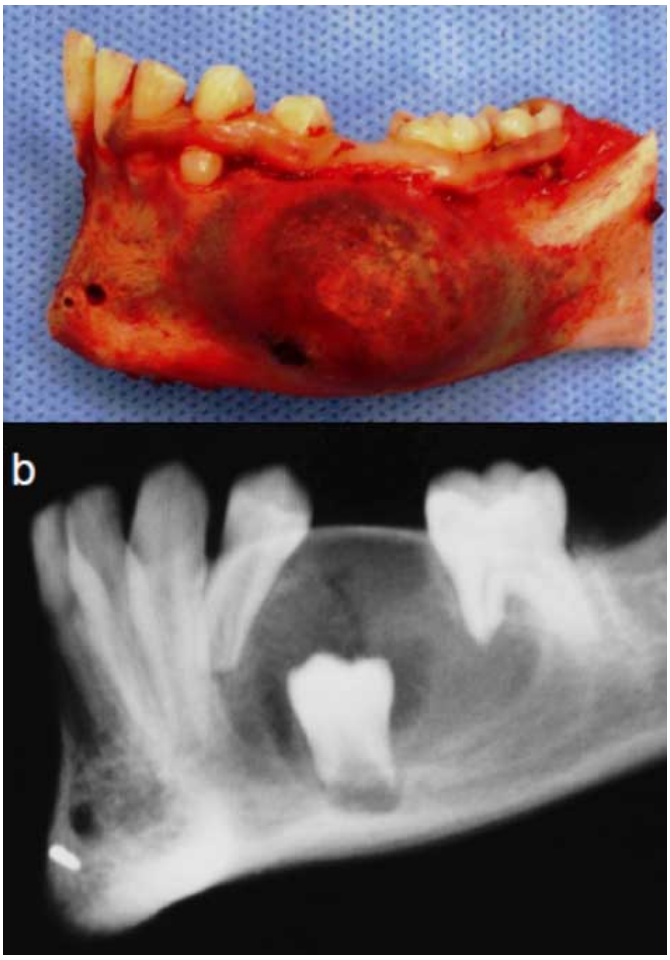

Fig. 6. a. Surgical specimen. b. intraoperative radiography.

Post-operative radiographic evaluation showed adequate resection margins (Fig. 7) and the final pathology was consistent with the initial diagnosis of solid/multicystic ameloblastoma. The patient has had an uneventful recovery and at this time she is being observed for spontaneous regeneration of the mandible and will be scheduled for reconstructive surgery in the future. She will be followed every three months for the first year, and then twice a year for the next 4 years.

DISCUSSION

Ameloblastoma is uncommon in children. The most commonly cited article is a review of 1.036 cases, where the average age was 38.9 years with only 2.2% (19 of 858) under 10 years and 8.7% (75 of 858) between 10 and 19 (Small & Waldron). However, this report was published in 1955 when adenoameloblastoma and ameloblastic fibroma were considered ameloblastomas. The first report of ameloblastoma in children was in 1962, where 7 cases were reported in children under the age of 9 years old but 2 of these cases were ameloblastic fibromas and 1 case odontoameloblastoma (Young & Robinson, 1962).

Ord *et al*. (2002) made a review of reported cases of ameloblastomas in children from 1970 to

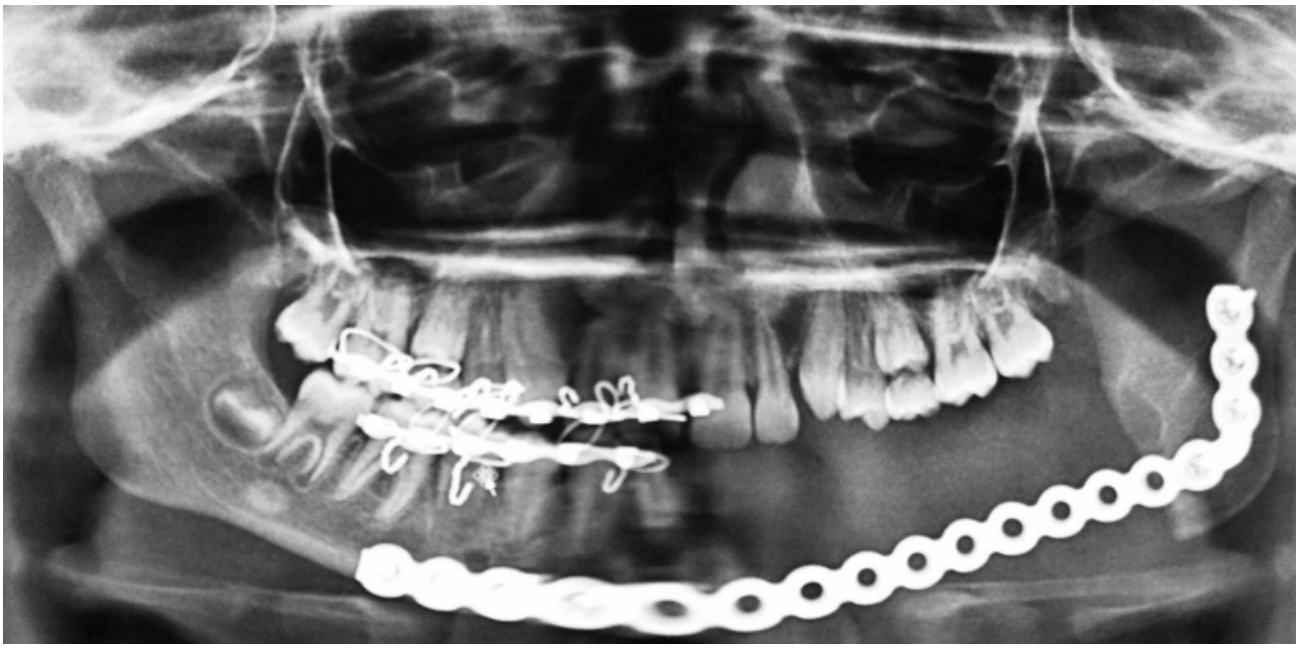

Fig. 7. Panorex, 1 day post-operative.

2001, comparing Western and African reports. This review showed an average age of 14.3 years (Western) and 14.7 years (African) and confirms that less than 10% of cases occur in children under 10-years-old. In adults, the gender ratio is 1:1. In Western children the ratio is 1:1.2 for male/female. While in African children, there is a male predominance of 1.4:1. The mandible is the most affected in adult ameloblastomas (85%), the third molar region being the most common site. This pattern is reflected in Western children where the angle of the mandible is affected 72% and the mandibular symphysis 5.8%. In contrast, African children were affected in the angle of the mandible 27.3% and in the mandibular symphysis 44.2%. The unicystic ameloblastoma variant is more common in Western children (76.5%). The solid/multicystic ameloblastoma is rare in children but in African children is the most prevalent (59.8%) assimilating the adult pattern.

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 (Robinson & Martinez, 1977; Shteyer *et al*., 1978). This radiographical similarity with dentigerous cyst would lead to initial treatment, marsupialization, curettage or enucleation. And only when the specimen is analyzed completely, definitive diagnosis is made and at that point one it should be determined if another treatment is necessary.

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 (Chapelle *et al*.).

In the treatment of children some authors recommend enucleation or only minimal treatment (Rapidis *et al.*, 1982; Isaacsson *et al*., 1986), while Fung *et al.* (1978) suggests that due to the greater amount of cancellous bone in young patients, the lesion would have a more aggressive course with more destruction, making the surgical procedure more difficult and demanding.

Ameloblastoma management remains controversial, but 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. However, some recurrences have been observed beyond 10 years after initial treatment (Dunfee *et al*.).

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RESUMEN: El ameloblastoma según la clasificación de tumores odontogénicos de la OMS del 2005 lo clasifica como una neoplasia benigna de origen epitelial odontogénico. Compromete el 1-3% de neoplasias y quistes maxilares. El tumor es agresivo localmente, pero muchas veces asintomático; presenta un lento crecimiento que se manifiesta como un aumento de volumen facial o un hallazgo incidental radiográfico. Al examen clínico el tumor puede causar síntomas como dolor, ulceración, reabsorción radicular con movilidad dentaria y maloclusión. El ameloblastoma posee gran tasa de recurrencia si no es totalmente removido. Se presenta en casi todos los grupos etarios pero principalmente se diagnostica en la tercera o cuarta década de vida, el tumor es muy poco común en niños. El tratamiento del ameloblastoma es controversial y debido a la distinta incidencia y comportamiento en niños, hace las consideraciones quirúrgicas diferentes a los adultos. Por lo que presentamos un inusual caso de un ameloblastoma solido/multiquístico mandibular en una niña de 10 años. Además de una breve revisión de la literatura sobre casos reportados de esta patología en niños.

PALABRAS CLAVE: ameloblastoma solido, ameloblastoma multiquístico, tumores odontogénicos, niños.

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