



## Case Report

# A Rare Case of a Multicystic Ameloblastoma of the Lower Anterior Jaw in a 10-Year-Old Child

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### Abstract

**Background:** Ameloblastoma is a benign neoplasm of odontogenic epithelial origin. One to three percent of tumors and cysts of the jaws are comprised of ameloblastomas. Its occurrence is considered a rarity in the younger age group. We present an unusual case of a large ameloblastoma of the anterior mandible in a 10-year-old child. **Patient and Method:** A 10-year-old male was referred to the department of oral and maxillofacial surgery in relation to a large (5.0 x 5.0 cm) swelling of the lower anterior jaw causing lower facial asymmetry. The patient suffered from difficulty in speech and eating due to a massive soft tissue overgrowth intra-orally occupying the anterior mandible and causing displacement of the anterior dentition. The segmental osteotomy of the entire anterior mandible including the lesion was done. The resulting defect was reconstructed using a 3D reconstructive titanium plate. **Results:** There was no postoperative infection or foreign body reaction during the 12-month follow up period. The lower facial asymmetry was restored and the patient was able to restore his speech and eating ability. **Conclusion:** The age, size, type and anatomic location of the tumour make it a rarity! To the best of our knowledge, this is the first report on the occurrence of a large, multicystic, ameloblastoma affecting the anterior lower jaw of a 10-year-old child.

**Keywords:** Ameloblastoma; Custom-made implant; 3D Printing; Mandible reconstruction

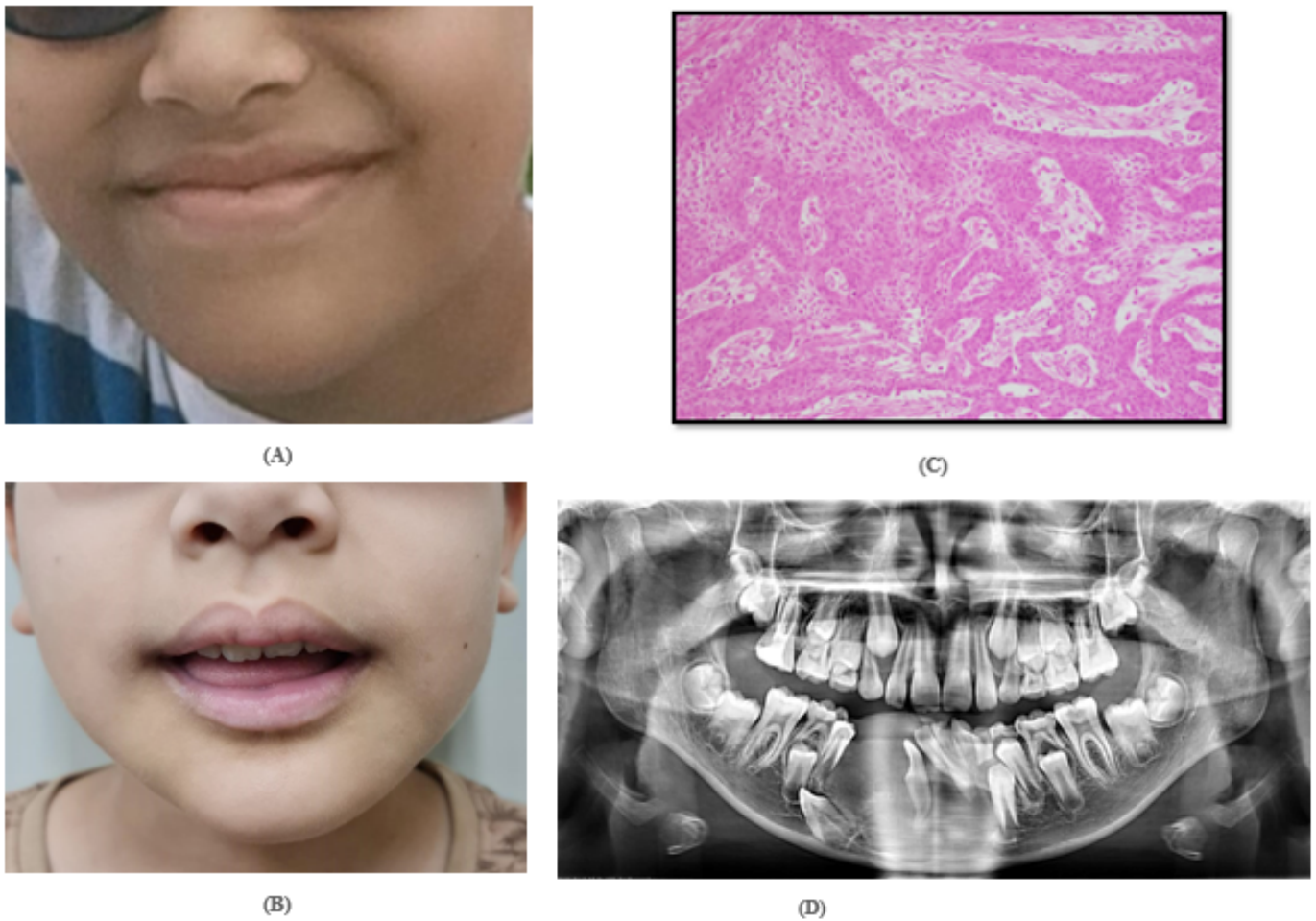
### Introduction

Ameloblastoma is an aggressive, benign, epithelial odontogenic tumor that comprises 10% of the odontogenic tumors [1]. Histologically, the tumor is classified into a diversity of subtypes of which conventional (multi-cystic) and uni-cystic ameloblastomas are the most frequent [2]. They primarily occur in adults, with a minority occurring in the pediatric population (aged 18 and younger) [2].

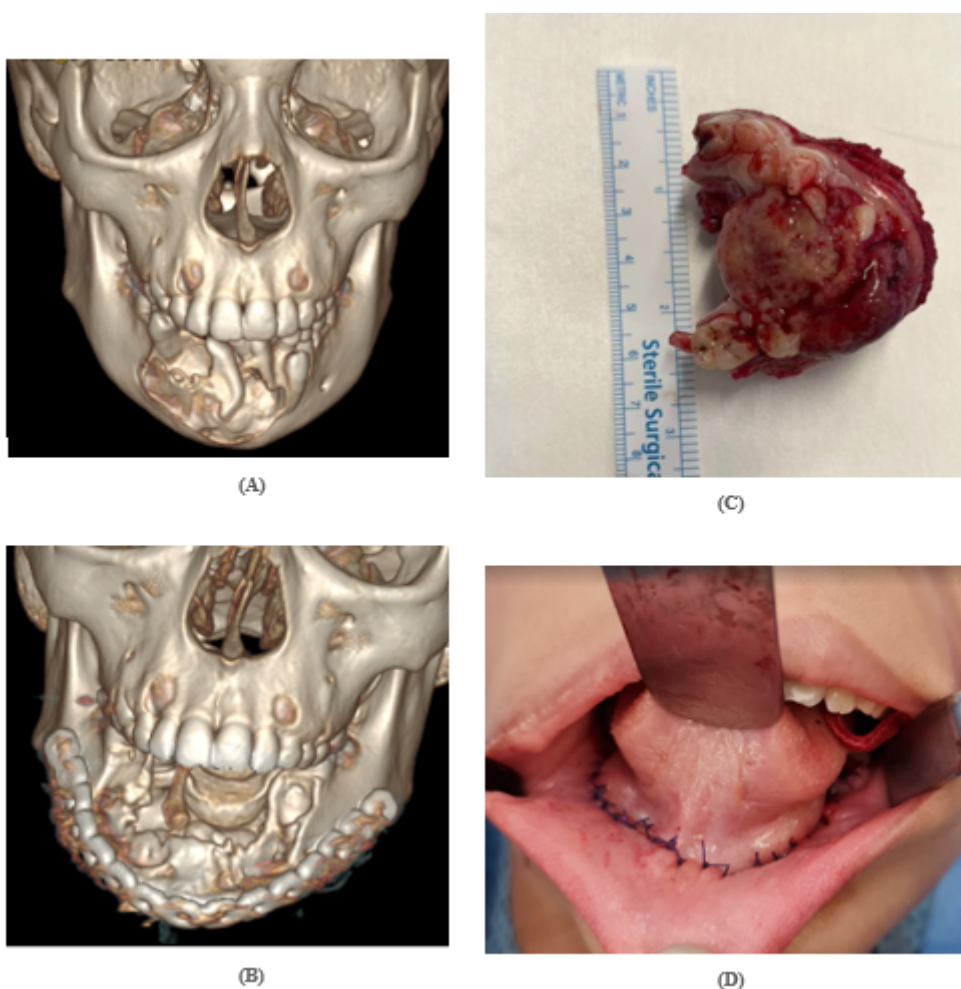
Ameloblastoma can theoretically arise from remnants of the dental lamina, enamel organ of developing tooth, the epithelial lining of odontogenic cyst or basal cells of the oral mucosa [3]. It occurs in almost all age groups, but mainly diagnosed in the third or fourth decade of life. Most cases (66%) affect the posterior mandible and ramus [4]. Ameloblastomas are usually asymptomatic and present as a slowly 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 [5]. This study presents an unusual case of a large follicular ameloblastoma of the anterior mandible in a 10-year-old child.

## Case Report

A 10-year-old male reported to the Department of Oral and maxillofacial surgery with an ulcerated large lump affecting the anterior mandible, which appeared two-years ago, as a small soft tissue peduncle. A dentist removed the soft tissue lesion without submitting it to histopathology. 12-months later, the lesion started to grow rapidly. Intra-oral examination indicated the presence of a large ulcerated swelling affecting the anterior mandible, measuring approximately 5.0 x 5.0 cm, and causing displacement of anterior dentition (Figure 1). A panoramic radiograph, performed by an oral and maxillofacial surgeon, showed multilocular radiolucency causing destruction of the entire anterior mandible (Figure 1). The case was misdiagnosed as a papilloma lesion. The histopathology outcome did not match the clinical and radiological picture. Hence, the biopsy had to be repeated at our department, which confirmed the diagnosis of a follicular multicystic ameloblastoma (Figure 1). The treatment plan included the resection of anterior mandible and reconstruction using 3D-reconstructive plate (implant) (Figure 2). The patient was offered to reconstruct the bony defect using a micro-vascular Fibular flap. But the parents denied this treatment modality. 12-months regular follow up, showed no postoperative infection or foreign body reaction. The lower facial asymmetry was restored and the patient was able to restore his speech and eating ability.



**Figure 1:** A) Pre-operative photo of the patient B) Post-operative photo of the patient C) Histopathology of the case showing anastomosing cords of odontogenic epithelium in a fibrous stroma. Some cords contain stellate reticulum-like cells centrally (H.E.; orig. magn. x 200) D) Panoramic view of the multicystic ameloblastoma affecting the entire lower jaw.



**Figure 2:** A) Pre-operative photo 3D CT scan showing the lesion occupying the anterior lower jaw B) Post-operative 3D CT scan showing the 3D implant used to reconstruct the bony defect C) En bloc resection of the excised lesion B) Intra oral view following closure of the soft tissue.

## Discussion

Ameloblastoma is a locally aggressive and infiltrative benign odontogenic neoplasm, which has a rare capacity to metastasize [6]. Ameloblastoma typically occurs within the third and fourth decade of life and is considered a rarity in the pediatric patient population, accounting for approximately 10-15% of all reported cases [6]. A rare case of a large multicystic ameloblastoma of the lower anterior jaw was presented here. The patient was a 10-year-old child. The age, size and anatomic location of the tumor make it a rarity!

Multicystic ameloblastoma has a classic radiographic presentation 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 un-erupted tooth. Most frequently, it is the mandibular third molar area, which is involved [7]. However, the multicystic ameloblastoma may appear radiographically as a unilocular lesion resembling other cystic lesions [5].

Histologically, most ameloblastomas have the follicular or plexiform pattern, although basaloid, granular cell or desmoplastic variations may also be seen [8]. It is generally accepted that there is no relationship between the individual patterns and the behavior of the tumor or its prognosis [9]. For this reason, pathologists may choose not to report the histologic pattern. The case presented showed features of follicular variant, possessing islands and anastomosing strands of odontogenic epithelium enmeshed in a fibrous stroma. These islands and strands contain basal cells that are columnar, hyperchromatic and lined up in a palisaded fashion at the periphery. The lesion is cytologically bland with no pleomorphism or mitotic figures noted [9].

Ameloblastomas are classified into four groups: Conventional (multicystic), unicystic, extra-osseous or peripheral and metastasizing types [6]. The multicystic ameloblastoma, is a benign epithelial odontogenic tumor of the jaw that is slow-growing, locally aggressive. Patients may present with a slow-growing mass, malocclusion, loose teeth, or more rarely paresthesia and pain; however, many lesions are detected incidentally on radiographic studies in asymptomatic patients [9]. The lesions usually progress slowly, but are locally invasive and will infiltrate through the medullary spaces and can erode cortical bone. If left untreated, they can resorb the cortical plate and extend into adjacent tissue [10].

Ameloblastomas are usually treated by segmental or marginal resection or conservatively (with marsupialization or enucleation with peripheral osteotomy with or without adjunctive therapy). Treatment of an ameloblastoma in adults generally requires resection to counter the aggressive nature of this tumour, particularly for the solid and multicystic type [6]. In children and adolescents, however, treatment of ameloblastomas is complicated by three factors:

1. The continuing facial growth and different bone physiology (more cancellous bone, increased bone turnover, and reactive periosteum), and the presence of unerupted teeth
2. The difficulty in the initial diagnosis and
3. Predominance of the unicystic ameloblastoma.

All these factors pose some special problems owing to concerns of the effects of resection and reconstruction on maxillofacial development [6]. Though there is consensus that ameloblastoma has to be treated aggressively to avoid recurrence, there is a dilemma on the applicability of an initial radical, extensive surgery in children [6].

The occurrence of ameloblastoma in children alters craniofacial growth and development; and often results in psychological and social disorders [6]. Treatment planning for ameloblastomas in children must take into consideration the age of the patient, type and size of the lesion, and reconstruction of the defect, thereafter. Treatment of ameloblastomas in pediatric patients is aimed at not only restoring function and aesthetics but must also consider the potential impact of the treatment and reconstruction on craniofacial growth. In addition, consideration must be given to whether the dissection is going to be sub-periosteal or supra-periosteal; together with the post-ablative grafting options in cases requiring resections. With sub-periosteal dissection, the periosteum is left intact with only the pathological lesion removed, and thus increasing the body's ability to form new bone especially in children [6]. The case discussed here was very unusual, and misdiagnosed for almost two years, which resulted in a large lesion (5.0 x 5.0 cm) destroying the entire anterior mandible. Conservative management was not an option due to the delay in the diagnosis. Segmental osteotomy of the entire anterior mandible including the lesion was done. The resulting defect was reconstructed using a 3D titanium implant. The aesthetic outcome was very good enabling the patient to restore his speech and eating ability.

## Conclusion

The age, size, type and anatomic location of the tumour make it a rarity! To the best of our knowledge, this is the first report on the occurrence of a large, multicystic, ameloblastoma affecting the anterior lower jaw of a 10-year-old child.

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