

## **Mandibular Ameloblastoma with Follicular, Plexiform and Cystic Components in a 14-Year-Old Patient: A Case Report**

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### **Abstract**

**Background:** Ameloblastoma is a rare benign odontogenic tumor characterized by slow but locally aggressive growth and a high risk of recurrence if not adequately treated. Although more frequently diagnosed in adults, it may occasionally occur in children and adolescents, where it can mimic odontogenic cysts, making the diagnosis particularly challenging.

**Case Presentation:** We report the case of a 14-year-old male who presented with a painless swelling on the right side of the mandible evolving over two months. Clinical examination revealed a firm swelling in the posterior mandibular region without mucosal changes. Panoramic radiography demonstrated a well-circumscribed unilocular radiolucency extending from the second right mandibular molar to the mandibular angle, encompassing the developing germ of the third molar, with cortical expansion but no root resorption. Aspiration of the lesion yielded yellowish citrine fluid. Surgical biopsy and histopathological analysis revealed islands of odontogenic epithelium with follicular architecture, areas of plexiform arrangement, and cystic degeneration, consistent with a follicular ameloblastoma with cystic and plexiform components. No signs of malignancy were observed.

**Conclusion:** Pediatric ameloblastomas are uncommon but should be considered in the differential diagnosis of mandibular radiolucent lesions associated with impacted teeth. This case highlights the importance of combining clinical, radiological, and histopathological evaluation to establish the diagnosis and guide conservative surgical management, followed by long-term surveillance to minimize the risk of recurrence.

**Keywords:** Ameloblastoma; Follicular; Plexiform; Cystic; Mandible

### **Introduction**

Ameloblastoma is an uncommon benign odontogenic tumor that originates from epithelial remnants, including the dental lamina, enamel organ, or the epithelial lining of dentigerous cysts, and is characterized by diverse radiological and clinicopathological presentations [1]. Clinically, it usually manifests as a slow-growing, asymptomatic swelling, often associated with cortical bone expansion or perforation. Without treatment, these lesions may reach significant size, resulting in facial deformity. This neoplasm accounts for approximately 1% of all jaw tumors and cysts, representing 13 - 78% of odontogenic tumors, with higher prevalence reported in regions such as Africa, India, and China [2]. It exhibits a locally destructive and infiltrative nature, with a high recurrence rate if not adequately managed [3]. The

mandible is affected about five times more frequently than the maxilla, particularly the molar-ramus region, which accounts for nearly 80% of cases [4].

According to the fifth edition of the World Health Organization (WHO) Classification of Head and Neck Tumors (2022), ameloblastomas are divided into five subtypes: extraosseous/peripheral ameloblastoma, metastasizing ameloblastoma, unicystic ameloblastoma, conventional ameloblastoma, and adenoid ameloblastoma, based on their anatomical location, histopathological features, and biological behavior [5]. Mixed histological presentations with overlapping features are not unusual and may complicate both diagnosis and prognosis. Unicystic ameloblastoma, frequently associated with impacted teeth, is more common in younger patients and generally carries a more favorable prognosis compared with conventional solid ameloblastomas.

Given its aggressive nature and risk of recurrence, ameloblastoma in pediatric patients requires early diagnosis and careful management to balance complete tumor eradication with preservation of mandibular growth and function. The present case describes a mandibular ameloblastoma in a 14-year-old male, exhibiting a combination of follicular, plexiform, and cystic components. The rarity of such mixed histopathological features in a pediatric patient highlights the importance of comprehensive diagnostic work-up and emphasizes the need for long-term follow-up in order to minimize the risk of recurrence.

### Case Presentation

A 14-year-old male was referred by his dentist to the Oral Surgery Department of the Dental Consultation and Treatment Center of Casablanca for evaluation of a painless swelling in the right mandibular angle region, first noticed two months earlier by his mother. The swelling was progressively enlarging, firm on palpation, and not associated with pain, paresthesia, or signs of infection. The patient was otherwise healthy, with no significant medical or family history.

Clinical examination revealed a slight facial asymmetry due to swelling in the right mandibular body-angle region (Figure 1). Intraorally, vestibular cortical expansion was observed extending from the second right mandibular molar to the angle of the mandible. The overlying mucosa appeared intact, with no ulceration or inflammatory changes. Aspiration of the lesion produced a yellowish citrine fluid (Figure 2).



**Figure 1:** Extraoral view showing slight facial asymmetry due to swelling in the right mandibular angle region.



**Figure 2:** Aspiration of the lesion yielding a yellowish citrine fluid.

Panoramic radiography showed a well-defined unilocular radiolucency extending from the distal root of the second right molar to the mandibular angle, encompassing the developing germ of wisdom tooth. Cone-beam computed tomography (CBCT) confirmed a well-demarcated unilocular radiolucent lesion in the right posterior mandible, extending from the distal aspect of 47 to the mandibular angle and surrounding the follicle of the developing third molar, with no cortical integrity left anteriorly and a thin cortical posteriorly. The inferior alveolar canal was displaced inferiorly but remained intact (Figure 3). Radiological findings suggested an odontogenic lesion, with differential diagnoses including unicystic ameloblastoma, odontogenic keratocyst, and dentigerous cyst.



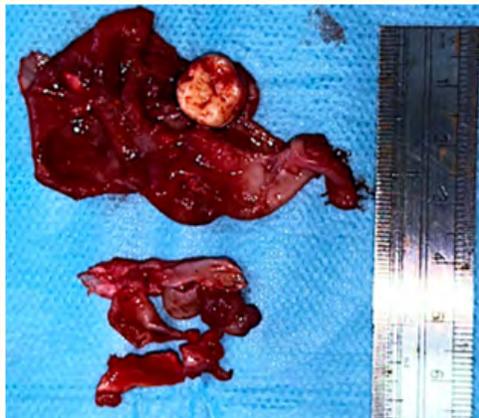


**Figure 3:** Pre-operative radiographs; A: Panoramic radiograph, B: CBTC.

Given the increased risk of pathological mandibular fracture, surgical intervention was indicated. After obtaining informed consent from the patient’s mother, the lesion was enucleated with curettage under local anesthesia, together with extraction of the developing third molar. A full-thickness mucoperiosteal flap was raised in the right mandibular molar region (Figure 4). During deep curettage, the patient experienced brief electric-shock-like sensations, suggesting proximity to the inferior alveolar nerve. The surgical specimen, including the lesion and the third molar germ, was submitted for histopathological examination (Figure 5).

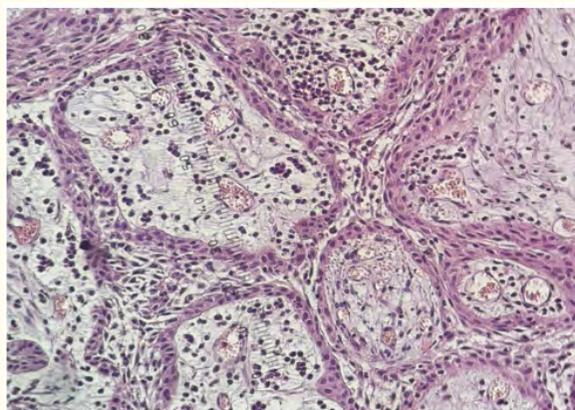


**Figure 4:** Intraoperative view after flap elevation.



**Figure 5:** Surgical specimen including the lesion and the developing third molar germ.

Microscopic examination revealed islands of odontogenic epithelium with peripheral palisading of columnar cells exhibiting reverse nuclear polarity, consistent with a follicular pattern. Areas of interlacing cords and trabeculae indicated a plexiform architecture, while cystic degeneration of the epithelial lining was also observed. The stroma showed focal calcifications. No cellular atypia or abnormal mitotic activity was detected. These features confirmed the diagnosis of conventional ameloblastoma with follicular, plexiform, and cystic components (Figure 6).



**Figure 6:** Photomicrograph showing follicular and plexiform patterns with cystic changes (H&E stain, magnification X40).

The immediate postoperative course was initially uneventful; however, after one week, the patient developed signs of localized infection at the surgical site (Figure 7). Empirical antibiotic therapy was prescribed, resulting in complete resolution within a few days. At the three-month follow-up, clinical and radiographic assessment showed satisfactory healing with no evidence of recurrence (Figure 8). At eight months, panoramic radiography revealed bone regeneration at the surgical site, and the patient remained asymptomatic (Figure 9). Long-term clinical and radiographic surveillance is ongoing, given the high recurrence potential of ameloblastomas.



**Figure 7:** Post-operative follow-up.



**Figure 8:** Panoramic radiograph at three months postoperative showing satisfactory bone healing.



**Figure 9:** Panoramic radiograph at eight months postoperative demonstrating bone regeneration at the surgical site.

## Discussion

Ameloblastoma represents the second most common odontogenic tumor, accounting for approximately 1% of all jaw tumors and cysts [6]. It occurs predominantly in the mandible, particularly in the molar-ramus region, with up to 80% of cases arising at this site, and is most often diagnosed between the ages of 30 and 60 years [7,8]. In a large systematic review and meta-analysis including 6,446 patients, Hendra, *et al.* (2019) estimated the global incidence of ameloblastoma at 0.92 per million person-years (95% CI: 0.57-1.49). Solid/multicystic types accounted for 67.7% of cases, while follicular (24.8%) and plexiform (24.7%) patterns were the most frequent histopathological variants [9].

Although well-characterized clinically and histologically, the exact etiological factors remain incompletely understood. Advances in molecular biology have highlighted the role of genetic alterations in its pathogenesis. Among these, the BRAF V600E mutation, reported in 63-82% of ameloblastomas, has emerged as a key driver [10,11]. This mutation activates the Mitogen-Activated Protein Kinase (MAPK) pathway, specifically the Extracellular signal-Regulated Kinase (ERK) cascade, leading to uncontrolled cell proliferation, enhanced survival, and local tissue invasion. In addition to BRAF, other molecular alterations have been described, including overexpression of NOTCH4, aberrant activation of the Sonic Hedgehog (SHH) pathway, and dysregulation of bone remodeling through the RANK/OPG system (Receptor Activator of Nuclear Factor- $\kappa$ B and Osteoprotegerin) [12].

Pediatric cases remain rare. In children and adolescents, clinical and radiographic features frequently mimic odontogenic cysts, such as dentigerous cysts or odontogenic keratocysts, which may delay or complicate diagnosis. In a retrospective series of 76 cases, Da Cruz, *et al.* (2022) reported significant radiographic differences between pediatric and adult patients [13]. Among younger patients (22.4% of cases), lesions were more frequently well-circumscribed, unilocular, and associated with an unerupted tooth (64.7%,  $p < 0.001$ ), whereas adults more often presented with multilocular lesions and ill-defined margins (27.1%,  $p = 0.01$ ). Despite comparable histopathological features across age groups, these radiographic differences underscore the diagnostic challenge in children, as such presentations may closely resemble odontogenic cysts. Our case, involving a well-defined unilocular lesion associated with the follicle of an unerupted third molar, aligns with this pediatric radiographic profile. The radiological appearance of ameloblastomas in children may reflect both the biological behavior of the tumor and host-related factors. Pediatric jawbones differ from adults in structure, being characterized by a higher proportion of bone marrow, increased cellularity, and active bone remodeling related to growth and tooth eruption.

Our findings are consistent with the case described by Urechescu, *et al.* (2024), who reported a 16-year-old female with a mandibular ameloblastoma presenting follicular, plexiform, cystic, and acanthomatous components [14]. Similar to our patient, their lesion radiographically mimicked an odontogenic cyst and was managed by enucleation and curettage. However, delayed consultation during the COVID-19 pandemic led to a larger lesion with cortical perforation and root resorption, underscoring the importance of early detection and timely intervention in pediatric cases. A further comparison can be drawn with the case reported by Kajla, *et al.* (2022), involving a 43-year-old female with a mandibular ameloblastoma characterized by follicular and plexiform patterns [15]. In contrast with our patient, that lesion was multilocular, extensive, and required segmental resection with reconstruction. Together, these reports illustrate the wide spectrum of clinical presentations and histopathological variants of ameloblastoma, as well as the influence of age and timing of diagnosis on treatment strategy.

Histopathologically, ameloblastomas display several growth patterns, with follicular and plexiform variants being the most frequent [16]. Mixed presentations with cystic degeneration, as observed in our case, are not uncommon and may further complicate both diagnosis and prognosis. In the present patient, however, histology confirmed a conventional ameloblastoma combining follicular, plexiform, and cystic components, which represents an uncommon presentation in this age group.

The optimal surgical approach for ameloblastoma remains a matter of ongoing debate. A systematic review and meta-analysis by Hendra, *et al.* (2019) demonstrated significantly lower recurrence rates following radical surgery-8% (95% CI: 4-13) for solid/multicystic lesions and 3% (95% CI: 1-7) for unicystic forms-compared with 41% (95% CI: 34-48) and 21% (95% CI: 16-26), respectively, after conservative treatment [17]. Similarly, Qiao, *et al.* (2021), in a meta-analysis of 942 cases, reported recurrence rates of 12% (95% CI: 9-16) after aggressive surgery versus 30% (95% CI: 23-39) after conservative management, with multicystic variants showing the highest recurrence risk (38%, 95% CI: 32-46) [18]. These findings highlight the superior oncological safety of radical resection. However, in pediatric patients, the choice of treatment must balance complete tumor eradication with preservation of mandibular growth and function. Conservative approaches, while associated with a higher recurrence risk, may be justified in this context. In the present case, conservative management with enucleation and curettage was selected due to the patient's young age, emphasizing the importance of long-term surveillance.

Although a localized postoperative infection developed at one week, it resolved promptly with antibiotic therapy, and the patient showed satisfactory healing with bone regeneration at the eight-month follow-up. Given the well-documented recurrence potential of conventional ameloblastomas-particularly those with follicular and plexiform components-strict, long-term clinical and radiographic follow-up is essential.

## Conclusion

Pediatric ameloblastomas represent a diagnostic challenge, as their clinical and radiographic features frequently resemble those of odontogenic cysts, particularly dentigerous cysts and odontogenic keratocysts. Therefore, accurate diagnosis requires meticulous correlation of clinical, radiological, and histopathological findings. Given their aggressive behavior and high risk of recurrence, pediatric ameloblastomas demand early recognition and carefully tailored management strategies that ensure complete tumor eradication while preserving mandibular growth and function.

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