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CASE REPORT

# Maxillary Ameloblastic Fibroma Mimicking Dentigerous Cyst in a Child: A Case Report

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**Purpose:** Ameloblastic fibroma (AF), an exceedingly rare benign mixed odontogenic tumor, scarcely occurs in the anterior region of the maxilla. This case report aims to delineate the formidable challenges faced during the early diagnosis of such cases, with the ultimate goal of averting missed diagnoses.

**Methods:** A 6-year-old female patient presented with the delayed eruption of bilateral maxillary central incisors. As early as 2 years old, tooth 62 was found to be impacted, accompanied by a supernumerary tooth in the anterior maxilla upon X-ray examination at another facility. Currently, for supernumerary tooth extraction, the child visited our hospital. Cone-beam Computed Tomography (CBCT) disclosed a supernumerary tooth between 11 and 21, along with a well-defined 5mm×6mm×6mm low-density shadow encircling the crown of impacted permanent tooth 62. The clinical diagnosis was anterior maxillary supernumerary tooth, impacted 62, and dentigerous cyst.

**Results:** Maxillary mass resection, extraction of impacted 62, and removal of the supernumerary tooth were carried out. Post-operative pathology confirmed AF, and long-term follow-up revealed no tumor recurrence.

**Conclusion:** Despite its rarity, AF should be contemplated in pediatric patients with tooth eruption delay and radiographic low-density lesions with irregular edge. This case offers invaluable insights for clinicians in diagnosing and managing small, early-stage lesions. Probing into the causes of non-erupted deciduous teeth may contribute to early lesion detection.

**Keywords:** odontogenic tumor, ameloblastic fibroma, supernumerary teeth, maxilla, pediatric dentistry

## Introduction

This article presents a case of AF that mimicked a dentigerous cyst and occurred in the anterior region of the maxilla. Additionally, it conducts a comprehensive analysis of the clinical manifestations, imaging characteristics, and treatment approaches while integrating relevant literature for a more in-depth discussion. Such documentation carries significant clinical significance. Ameloblastic fibroma (AF) is a rare mixed odontogenic benign tumor, accounting for only 2% of odontogenic tumors.[1] It is characterized by the presence of neoplastic epithelial and ectomesenchymal tissues. Notably, this tumor does not induce the formation of enamel and dentin. Over 80% of cases occur in the mandible, especially in the posterior dental region, while it is relatively rare in the anterior region of the maxilla.[2] Due to its association with impacted teeth and a well-defined radiographic border, it is often misdiagnosed as a dentigerous cyst.

Currently, research on AF mainly focuses on common cases in the mandible. Systematic studies on lesions in rare locations such as the anterior region of the maxilla are relatively scarce. Moreover, existing literature reports on the

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clinical characteristics, diagnosis and treatment rules, and prognosis of AF in the pediatric population are also scattered, lacking support from large-sample data, resulting in obvious research gaps.

This article reports a case of ameloblastic fibroma occurring in the anterior region of the maxilla that mimics a dentigerous cyst. In addition, it conducts a comprehensive analysis of the clinical manifestations, imaging features, and treatment methods of this case, and carries out in-depth discussions combined with relevant literature. The detailed recording and analysis of such rare cases can not only enrich the clinical data of AF occurring in special locations, provide references for clinicians to identify such atypical lesions, reduce the occurrence of misdiagnosis and mistreatment, but also lay a foundation for subsequent research on AF in relevant rare locations and special populations, which has important clinical significance and scientific research value.

This study was approved by the Medical Ethics Committee of Jinan Stomatological Hospital and informed consent has been obtained from the child's family.

## Case Report

The patient was a 6-year-old female who visited our hospital with the chief complaint that the upper anterior teeth had not erupted. When the child was 2 years old, tooth 62 was found to be impacted, and an X-ray examination at another medical institution revealed a supernumerary tooth in the anterior maxilla (Figure 1).

Specialty examination: The maxillofacial region exhibits bilateral symmetry, with normal skin coloration. The lips are free of purpura. There is no significant tenderness anterior to the bilateral tragus. Both the degree of mouth opening and the opening pattern are within normal limits. Teeth 51 and 61 are retained, and tooth 62 has not erupted. The oral hygiene condition is good. There is no soft scale (-) and no dental calculus (-). There are no obvious abnormalities in the gingival mucosa. There is no redness or swelling at the orifices of the bilateral parotid gland ducts, and when squeezed, clear fluid is secreted. No enlarged lymph nodes are palpable in the maxillofacial and neck regions.

The cone-beam computed tomography (CBCT) examination showed that there was an inverted supernumerary tooth between teeth 11 and 21. Tooth 62 was impacted at a low position. There was a low-density radiolucent shadow surrounding the crown, with a size of approximately 5mm × 6mm × 6mm. The boundary was clear, but the margin was not regular. The root apexes of teeth 11 and 21 were at the 8th stage of development, and the periodontal membrane was continuous and intact. The root apex of tooth 22 was at the 7th stage of development (Figure 2). Based on the radiographic and clinical findings, a tentative diagnosis of dentigerous cyst was formulated.

Under general anesthesia combined with local anesthesia, the extraction of the supernumerary tooth and the removal of the mass in the maxilla were performed. During the operation, it was shown that the mass was cystic, with a thick wall, and no obvious contents were seen. The mass was sent for postoperative pathological examination.

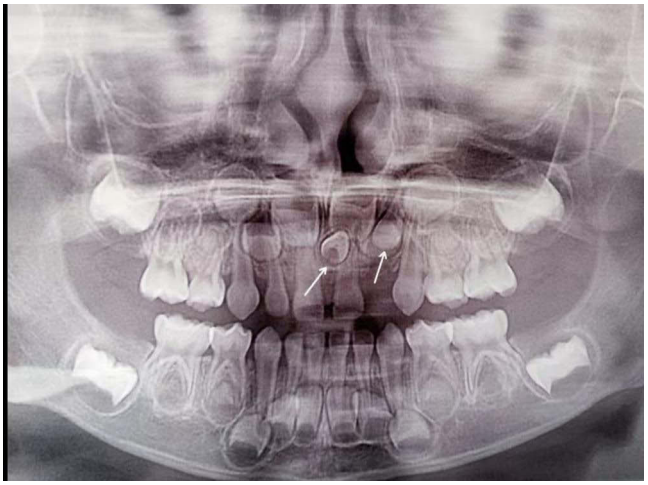

**Figure 1** The arrows indicate a supernumerary tooth and an impacted tooth 62 in the anterior maxillary region. Upon meticulous inspection, a low-density shadow is discernible encircling the coronal portion of tooth 62.

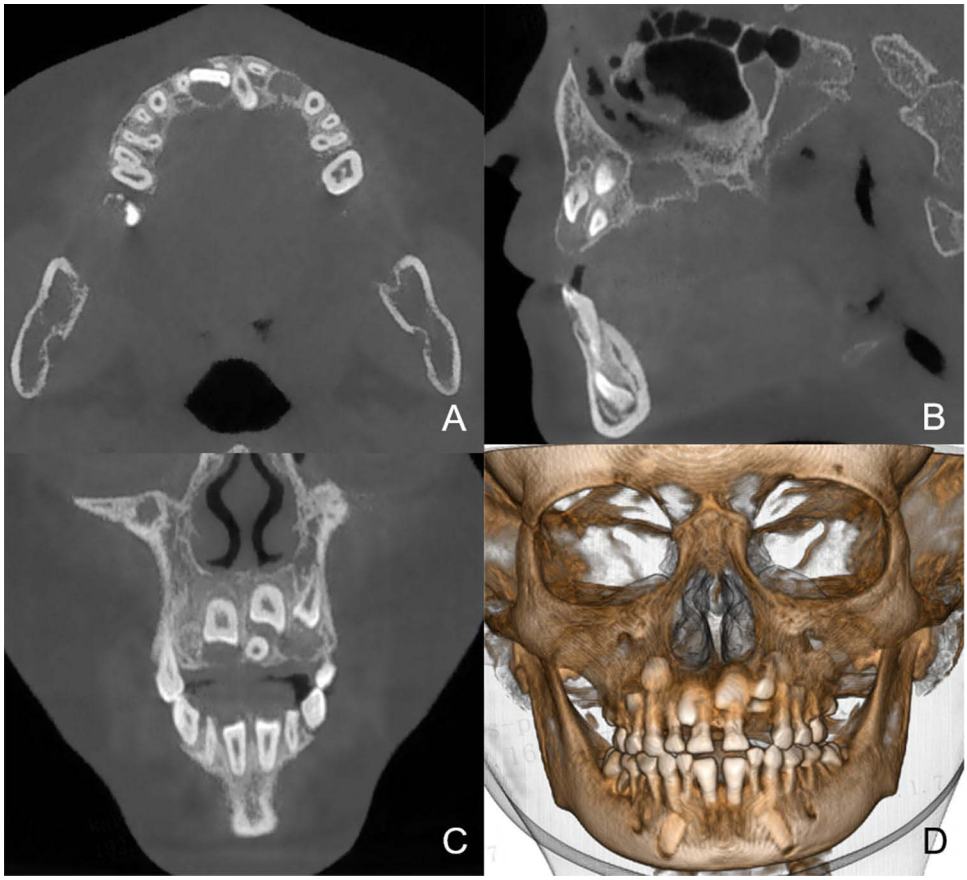


**Figure 2** Preoperative CBCT images (September 26th, 2023)(**A**) (axial view) (**B**) (sagittal view) (**C**) (coronal view) (**D**) (three-dimensional reconstruction) show a low-density radiolucent shadow surrounding the crown of tooth 62 and a supernumerary tooth in anterior maxillary region.

Pathological result: Tumor cells were arranged in the form of cords and islands. The periphery was composed of cuboidal or columnar cells, and there might be a small number of stellate reticulum cells in the center. Immunohistochemical results: CK-pan: (+), Vimentin: (+), CK5/6: (+), CK7: (+), S-100: scattered (+), Ki-67: (the number of positive cells was approximately 2%), P63: (+). The pathological diagnosis was ameloblastic fibroma (Figure 3).

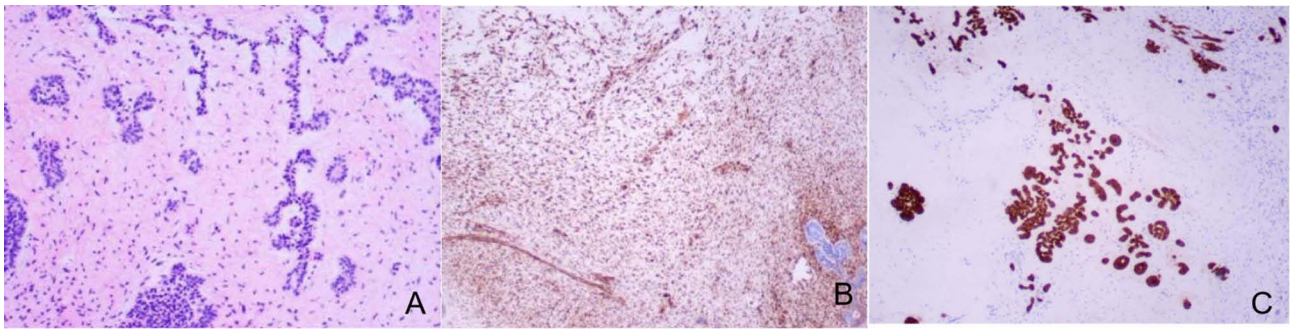


**Figure 3** Pathological diagnosis: Ameloblastic fibroma. (**A**) (Histopathological microscopy): The lesion consists of proliferative spindle cells and numerous nest-like basaloid cells. (**B** and **C**) (Immunohistochemistry): CK-pan (+), Vimentin (+), CK5/6 (+), CK7 (+), S-100 focally (+), Ki-67 (positive cells account for approximately 2%), and P63 (+). These findings are consistent with ameloblastic fibroma.


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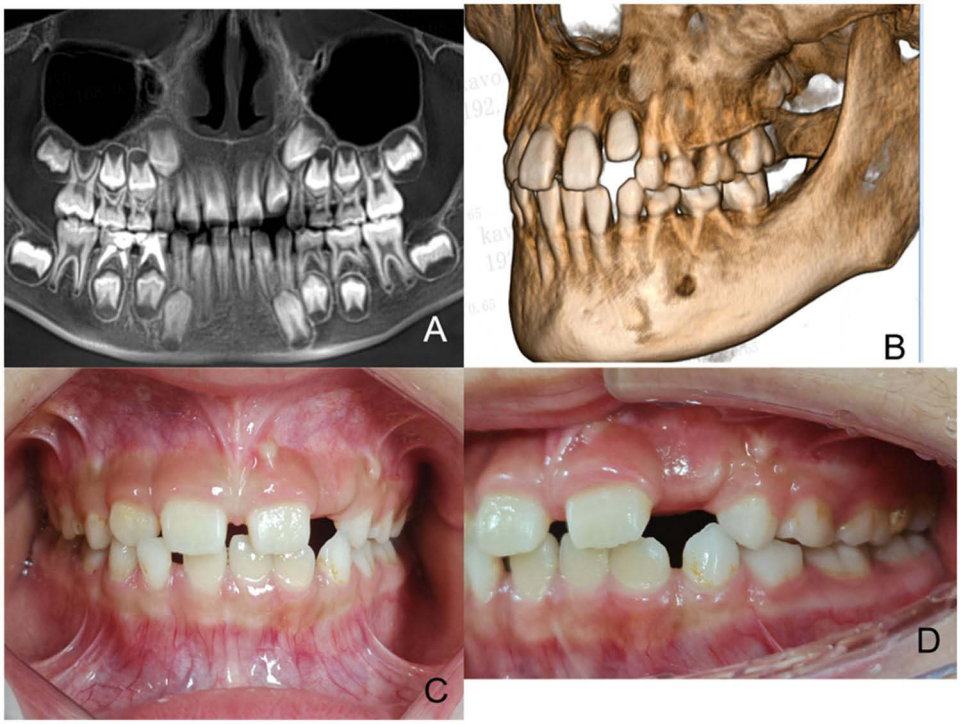


**Figure 4** Clinical and imaging manifestations: (**A** and **B**) (CBCT images) show that tooth 11 and tooth 21 have erupted normally, and the root apex of tooth 22 is at the 8th stage of development. (**C** and **D**) (Clinical examination) show that the teeth are normally developed.

The reexamination conducted on April 12th, 2025, revealed that teeth 11 and 21 had erupted normally. Additionally, the root apex of tooth 22 was found to be at the 8th stage of development, as illustrated in Figure 4. Notably, there were no indications of recurrence.

## Discussion

AF was first described by Kruse in 1891.[3] In 1946, AF was defined as a benign neoplastic entity and was named soft odontoma.[4] It was subsequently classified under mixed odontogenic neoplasms by the World Health Organization in 1992.[5] In 2022, AF was classified as a benign mixed epithelial and mesenchymal odontogenic tumours by the WHO.[6] AF is more commonly seen in children and adolescents, with an average age of onset of 15 years old (ranging from 7 weeks to 89 years old), and there is no obvious gender preference (some studies show a slight male predominance).[7,8] These reports were in accordance with our case, wherein the patient was 6 years old. In the early stage, AF usually has no obvious subjective symptoms. As the disease progresses, patients may experience local mass in the jawbone, swelling and bulging, tooth loosening, etc. Some patients are incidentally diagnosed during routine X-ray examinations. A few cases are also accompanied by symptoms such as pain and numbness of the lower lip. Our case is a small lesion with no obvious subjective symptoms. AF is often accompanied by the obstruction of tooth eruption. Our case is also associated with impacted tooth 62.

Pathological Features: The ameloblastic fibroma is composed of epithelial components arranged in an enamel organ-like pattern and a cellular, myxoid stroma. The cells exhibit no obvious atypia or nuclear polymorphism. The epithelial components typically manifest as cords or islands. The periphery consists of cuboidal or columnar cells, while the central area resembles the stellate reticulum layer. The stromal components predominantly consist of loosely arranged fibroblasts, and myxoid degeneration can sometimes be observed. Blum proposed the “maturation theory”, which suggests that an AF will develop through a continuum of differentiation and maturation into an ameloblastic fibroma odontoma and eventually to an odontoma. Accordingly, an AF would be an early stage of developing odontoma.[9,10] This theory is controversial. Some authors claimed that AF is a separate specific neoplastic entity that does not develop into a more

differentiated odontogenic tumor.[7] In the present case, thanks to the presence of a supernumerary tooth, there was an opportunity to observe the growth of the lesion over a four-year interval. The lesion did not exhibit evident maturational progression toward odontoma over a four-year period.

In this case, CBCT played a significant role. CBCT images clearly showed the location, extent and nature of the lesion. Radiographically, ameloblastic fibromas typically present as unilocular lesions; larger lesions may exhibit a multilocular appearance with smooth, well-defined margins.[2] AF can be radiographically confused with dentigerous cysts due to their common association with unerupted teeth. In this case, CBCT showed no obvious bone expansion, which is different from dentigerous cysts. The coronal view revealed that the margin of the lesion presented a lace-like appearance.

Histological examination is the gold standard for diagnosing AF. Microscopic Examination: The biopsy specimen reveals a tumor composed of neoplastic epithelium and primitive mesenchymal tissue, exhibiting typical features of a mixed odontogenic tumor. The epithelial component primarily consists of cord-like or island-like arrangements, with peripheral columnar cells demonstrating palisading nuclei and hyperchromasia, resembling ameloblasts. The central areas contain stellate reticulum-like cells with loose arrangements, similar to the enamel organ structure of a dental germ. However, no enamel or dentin formation is observed, consistent with the characteristic lack of inductive changes in ameloblastic fibroma (AF). The mesenchymal component is composed of densely packed spindle cells with scant cytoplasm and oval nuclei, interspersed with minimal collagen fibers, exhibiting features of immature connective tissue resembling dental papilla. The tumor is relatively well demarcated from surrounding normal tissue though focal areas show epithelial cords extending into the mesenchymal stroma. Immunohistochemistry: The epithelial component is positive for cytokeratin (CK), while the mesenchymal component expresses vimentin. These histopathological findings align with the typical features of AF as reported in the literature.

It is necessary to differentiate AF from diseases such as ameloblastoma, odontoma, and odontogenic fibroma. The epithelial components of ameloblastoma are mostly in a follicular or plexiform pattern, and the stroma is fibrous connective tissue; odontoma contains hard dental tissues; the odontogenic fibroma has fewer epithelial components, and the stroma is fibrous tissue.

Surgical resection is the main method for treating AF. The specific surgical approach needs to be selected according to the size and location of the tumor. For smaller tumors, curettage can be used; for larger tumors or recurrent tumors, partial resection of the jawbone is required. During the operation, it is necessary to ensure a sufficient resection range, and if necessary, segmental resection of the jawbone should be performed to reduce the risk of recurrence.[11]

The recurrence rates of AF reported in the literature vary considerably, with values ranging from 12.5% to 60%.[12,13] Multiple recurrences may lead to malignant transformation into ameloblastic fibrosarcoma (AFS). In our case, after one and a half years of follow-up, there were no signs of recurrence either radiographically or clinically, and the teeth erupt normally. Limitations of this study: One and a half years of follow-up period is relatively short, making it difficult to evaluate late recurrence; CBCT cannot assess soft tissues, so further MRI studies are recommended.

## Conclusion

For AF with cystic changes, a biopsy is mandatory for definitive diagnosis, while CBCT aids in differential diagnosis. Regarding management, small AF lesions require conservative resection, whereas extensive or recurrent cases necessitate radical surgery.[14] Given the high recurrence risk, lifelong postoperative monitoring is essential. This case provides insights into the "maturation theory" of AF. Due to the rarity of AF in pediatric patients and maxillary involvement, current evidence remains limited. Further studies are crucial to establish robust diagnostic and therapeutic protocols.

## Ethics Statement

This study is a retrospective case report and conforms to ethical norms. In accordance with the policies of the Ethics Committee of Jinan Stomatological Hospital, this retrospective study is exempt from ethical approval, including for publication, as it involves no intervention and ensures adequate privacy protection.

## Informed Consent

Informed consents to publish have been obtained from the patient's parents for the case.

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## Disclosure

The authors report no conflicts of interest in this work.

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