



Delayed tooth eruption associated with an ameloblastic fibro-odontoma

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Abstract

Delayed eruption of a single primary tooth is an uncommon event. Excluding a previous traumatic insult, the presence of a pericoronal odontogenic cyst or neoplasm is the primary cause for this abnormality. This case report describes the clinical and radiographic features of an ameloblastic fibro-odontoma in a young child, who presented with delayed eruption of the primary mandibular canine and prominent buccal expansion. A differential diagnosis for mixed, radiolucent and radiopaque lesions of the jaws will be discussed. (Pediatr Dent 23:253-254, 2001)

Although an uncommon event, odontogenic cysts and neoplasms are an important consideration when a single primary tooth fails to erupt. This is especially true for primary canines and molars because traumatic impaction rarely involves these teeth. Odontomas are the most common lesions associated with delayed eruption of these teeth, but when there is concurrent bony expansion, other odontogenic lesions are usually responsible for these signs.¹ This case report describes the clinical and radiographic features of an ameloblastic fibro-odontoma in a young child, in addition to an age-appropriate differential diagnosis.

Case history

A healthy, 31 month-old Hispanic boy was referred for evaluation of an unerupted primary canine of the left mandible. Recently, a bony hard mass at the site of the missing tooth became noticeable to the mother. Clinical examination revealed mild facial asymmetry of the lower one-third of the face. Intraorally, both buccal and lingual cortical expansion of the left mandible was observed with lingual displacement of the lateral incisor and distalization of the molars (Fig 1). Due to the size of the lesion the child was beginning to occlude on the bony swelling with the opposing teeth. Palpation of the area revealed an asymptomatic, hard enlargement with smooth contours that was covered by normal mucosa. Crepitus, lymphadenopathy or increased tissue warmth was not present. A periapical radiograph demonstrated a localized radiolucency with dilaceration of the mesial root of the first primary molar and posterior displacement of the developing premolar. A modified occlusal radiograph was most useful in demonstrating the pertinent features of this entity (Fig 2). This expansile, mixed radiolucent and radiopaque lesion surrounded the crown of an inferiorly displaced canine. Significant buccal expansion



Fig 1. Significant buccal expansion in the area of the unerupted, primary, mandibular canine.

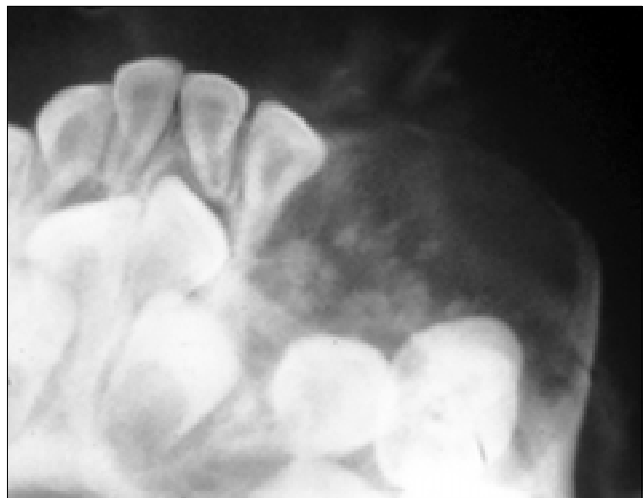


Fig 2. Modified occlusal radiograph demonstrating an expansile radiolucent and radiopaque lesion of the anterior mandible.

and thinning of the cortical plate of bone was well-illustrated by this film. Based on these findings, the child was referred for surgical excision of the jaw lesion under general anesthesia.

The histopathologic findings included a tumor with cords and islands of odontogenic epithelium, conglomerate foci of enamel and dentin and tiny rudimentary tooth-like structures

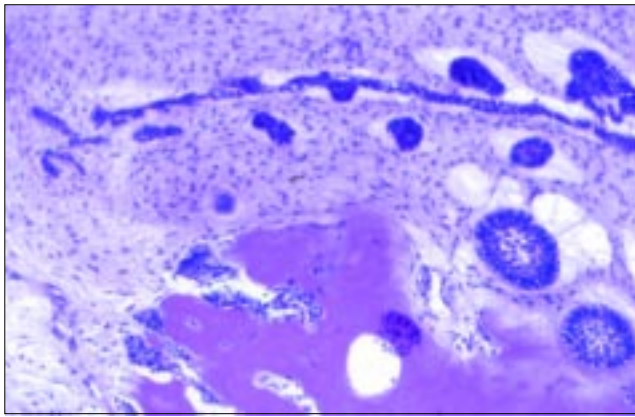


Fig 3. Photomicrograph of an ameloblastic fibro-odontoma (hematoxylin-eosin, magnification 200X).

(Fig 3). The loose and cellular connective tissue stroma resembled the dental papilla. In addition, a well-formed primary canine was intimately associated with the tumor. A diagnosis of ameloblastic fibro-odontoma was rendered based on these microscopic findings.

Discussion

The AF-O is an uncommon odontogenic tumor that resembles both an ameloblastic fibroma and complex odontoma. Most cases occur in children with the mean age ranging from 8 to 11 years-old but cases as young as 9 months-old have been reported.² The AF-O occurs equally in both jaws but favors the anterior region when it involves the maxilla, in contrast to the posterior region in the mandible. Typically a slow growing tumor, common clinical findings include a painless swelling and failure of one or more teeth to erupt. The radiographic appearance varies from a unilocular to multilocular radiolucency with well-defined margins and variable amounts of radiopaque material. The irregular or globular calcifications are usually located in the center of the lesion and are the same density as tooth structure. However, in very early lesions, mineralized product formation may not be evident on the radiographs. These lesions are often several centimeters in size and can result in significant displacement of the adjacent teeth.

The treatment of the AF-O usually involves conservative curettage, although large lesions may require a surgical splint to prevent fracturing of the jaw. In addition, an example of very rapid growth has been reported in a young child, following an incisional biopsy prior to definitive treatment.² Recurrences following conservative surgery are uncommon. The prognosis is excellent for this tumor; however, very rare cases of malignant transformation have been reported.³

Differential diagnosis

Other odontogenic lesions in children that present as a radiolucent lesion with irregular to globular radiopacities include the adenomatoid odontogenic tumor, the calcifying odontogenic cyst and the complex odontoma.⁴ The most common of these lesions is the complex odontoma, which is usually detected in the first 2 decades of life. Considered to be a developmental anomaly or hamartoma, the complex odontoma has a predilection for the pericoronal molar region. Most od-

ontomas are asymptomatic and are diagnosed because of delayed eruption of a tooth or as an incidental finding on a radiograph. Although most odontomas are described as a variably-sized calcified mass, exhibiting the same density as a tooth, and surrounded by a narrow radiolucent rim, in the young child the developing odontoma may be difficult to distinguish from the AF-O, both radiographically and microscopically.⁵ An important difference between these 2 odontogenic lesions is that the odontoma tends to be smaller and minimally expansile. Local curettage is the recommended treatment and recurrences are rare.

Similar to the odontoma, the adenomatoid odontogenic tumor (AOT) occurs predominately in children during the second decade and delayed tooth eruption is the alerting sign.³ This benign tumor has a distinct preference for the anterior jaws, in particular, the maxillary canine region. Although large lesions may exhibit asymptomatic buccal expansion, many AOTs are small and do not produce clinically significant distortion of the surrounding bone. Typically, the AOT presents as a well-delineated, unilocular radiolucency around the crown of a developing tooth. Fine flecks of calcification referred to as a "snowflake appearance" are a common finding. Extraction of the involved tooth and enucleation of the tumor are the recommended treatment. Rarely recurrences are documented.

The calcifying odontogenic cyst (COC) is an uncommon lesion that demonstrates variable clinical behavior.³ Although cystic, benign and malignant neoplastic subtypes are defined under this entity, when it occurs in young children, the majority are the cystic type associated with an odontoma. Similar to the AOT, this cystic lesion occurs usually in the anterior region of the jaws. A wide age range has been noted with this COC but there is a peak incidence in the second and third decades. The radiographic features of the COC include a unilocular or multilocular radiolucency that is associated with an unerupted tooth in a third of the cases. Approximately 50% of all COCs are associated with irregular or tooth-like opacities; however, if it is associated with an odontoma, calcifications are a constant feature. Asymptomatic expansion of the cortical bone, along with root resorption or divergence may be seen. In general, the COC occurring in this age group is treated by simple enucleation and recurrences are uncommon.

References

1. Otsuka Y, Mitomi T, Tomizawa M, Noda T: A review of clinical features in 13 cases of impacted primary teeth. *Int J Pediatr Dent* 11:57-63, 2001.
2. Baker WR, Swift JQ: Ameloblastic fibro-odontoma of the anterior maxilla. Report of a case. *Oral Surg Oral Med Oral Pathol* 76:294-7, 1993.
3. Waldron C: Odontogenic cysts and tumors. In *Oral and Maxillofacial Pathology*, BW Neville, DD Damm, CM Allen, JE Bouquot EDS. Philadelphia: WB Saunders Co, 1995, pp 493-540.
4. Steinberg MJ, Herrera AF, Frontera Y: Mixed radiographic lesion in the anterior maxilla in a 6-year-old boy. *J Oral Maxillofac Surg* 59:317-21, 2001.
5. Slootweg PJ: An analysis of the interrelationship of mixed odontogenic tumors – ameloblastic fibroma, ameloblastic fibro-odontoma, and the odontoma. *Oral Surg* 51:266-76, 1981.