Ameloblastic fibrodentinoma with a congenitally missing second premolar tooth: a case report

Hisazumi Ikeda, DDS, PhD,[a] Tokutarou Minamizato, DDS, PhD,[a] Shuichi Fujita, DDS, PhD,[b] and Izumi Asahina, DDS, PhD[a]
Nagasaki University Graduate School of Biomedical Sciences, Nagasaki, Japan

Ameloblastic fibrodentinoma (AFD) is a rare benign mixed odontogenic tumor that occurs predominantly in children and young adults. AFD is usually associated with delayed eruption of the tooth and shows painless swelling. We present a case of AFD affecting an 8-year-old Japanese girl with a complaint of delayed eruption of her left mandibular second premolar tooth. Intraoral examination did not reveal any sign of pain or swelling in the left mandible. Panoramic radiography revealed a radiolucent and well-circumscribed lesion, which contained radiopaque material at the premolar lesion of the left mandible. The second deciduous molar tooth was unerupted and the second premolar tooth was congenitally lacking. Our provisional diagnoses were odontogenic tumor. The biopsy confirmed AFD. The treatment included total excision of the tumor and the second premolar tooth. No recurrence has been noted during the 2 years of follow-up. (Oral Surg Oral Med Oral Pathol Oral Radiol 2014;117:e88-e91)

Although ameloblastic fibrodentinoma (AFD) has been defined as a neoplasm similar to ameloblastic fibroma (AF), it additionally exhibits inductive changes that lead to the formation of dentin.[1] AFD is an exceedingly rare histological variant of AF in which there is the formation of dentin or dentinoid tissue.[2] AFD arises mainly in the posterior mandible and is usually associated with unerupted molar teeth. Tumors related to the deciduous teeth usually arise in the incisor area, as they are related to the permanent teeth that develop in the molar region. AFD is more common in males than in females, and often grows slowly and is asymptomatic. Radiographically, it shows a fairly well delineated radiolucency with varying degrees of radiopacity.[3] The recommended treatment is conservative surgical excision.[4] At the present time, however, there is no agreement with regard to the preservation of the teeth affected by the tumor. The purpose of this article is to describe a patient who was followed up for 2 years after undergoing excisional surgery for a case of AFD that affected the left mandible body and included a congenital missing second premolar tooth.

CASE REPORT

In April 2010, an 8-year-old Japanese girl with delayed eruption of her left mandible second premolar tooth was referred by her general dentist to the Department of Oral and Maxillofacial Surgery at Nagasaki University Hospital. The medical and family histories were unremarkable. Clinical examination did not reveal any sign of pain or swelling in her left mandible. There was no history of local trauma or infection. She had good oral hygiene with extra- and intra-oral inspection. There was no visible swelling or expansion and the overlying mucosa appeared normal. Radiographic examination revealed well-defined radiolucency with radiopacity noted above the tooth crown of the unerupted deciduous second molar in the left mandible (Fig. 1). There was complete formation of the tooth located near the inferior border of her mandible. The patient also had a congenital lack of the second premolar tooth in her left mandible. Computed tomography revealed radiopaque masses in the well-circumscribed lesion in her left mandible (Figs. 2 and 3). The clinical diagnosis was benign odontogenic tumor, with suspicion of ameloblastoma.

The biopsy specimens revealed that the tumor consisted of odontogenic ectomesenchymal tissue and odontogenic epithelium arranged in islands, strands, and cords. The ectomesenchymal components showed moderate density of oval or spindle cells around the epithelial nests. A hyalinous change was noticed in the areas distant from the epithelial nests (Fig. 4). Some of the epithelial nests demonstrated the presence of an enamel organ-like structure with a peripheral palisading arrangement of the columnar cells (Fig. 5). In addition, a premature dentin-like matrix with enclosed cells was in contact with the odontogenic ectomesenchymal tissues. Enamel organ-like epithelial follicles and small epithelial nests were found adjacent to and within the dysplastic dentin matrix (Figs. 6 and 7). At the time of the biopsy examination, tooth germ-like structures with enamel and dentin formation were not seen, and there was no atypia noted in the tumor cells. After a histological diagnosis of AFD was made, the patient underwent surgical excision. The tumor was well demarcated and there was no perforation seen in the buccal and lingual cortex bone. Since the dental sac of the second premolar tooth adhered to the tumor, the tooth was also removed during the tumor excision. As the surgical specimens demonstrated similar histological features to the biopsy tissue, a final diagnosis of AFD was made. Radiographic observations

[a]Department of Regenerative Oral Surgery, Unit of Translational Medicine, Nagasaki University Graduate School of Biomedical Sciences.
[b]Department of Oral Pathology and Bone Metabolism, Unit of Basic Medical Sciences, Nagasaki University Graduate School of Biomedical Sciences.
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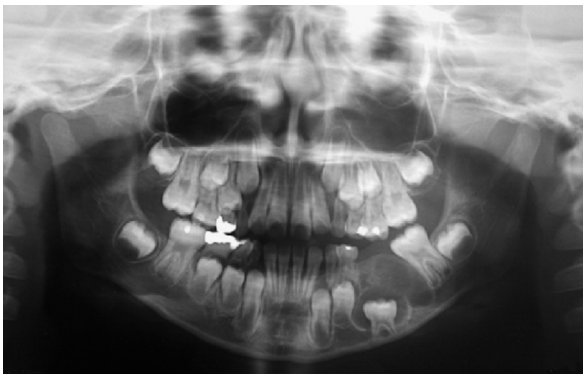

Fig. 1. Panoramic radiography revealed well-defined radiolucency with a radiopaque area above the tooth crown of the unerupted deciduous second molar in the left mandible.

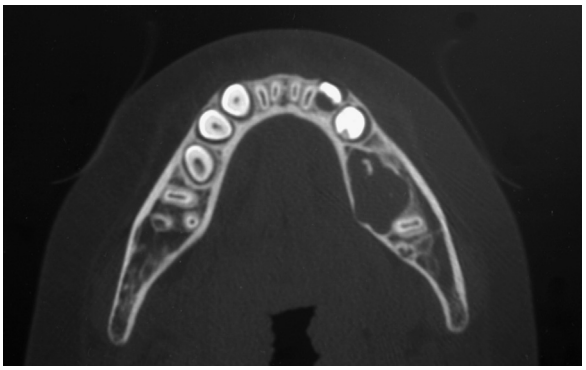

Fig. 2. Horizontal computed tomography showed calcified masses in the well-circumscribed lesion in the left mandible. Slight expansion and absorption were noticed in the lingual cortex in the left molar lesion.

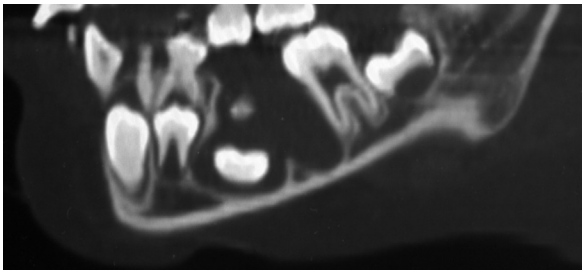

Fig. 3. Sagittal computed tomography showed calcified masses above the unerupted tooth in the well-circumscribed lesion.

after 12 months of follow-up indicated there was new bone formation (Fig. 8), and at 24 months after the surgical excision, there was no recurrence observed.

DISCUSSION

According to the World Health Organization,[5] AF, AFD, and ameloblastic fibroodontoma (AFO) are all

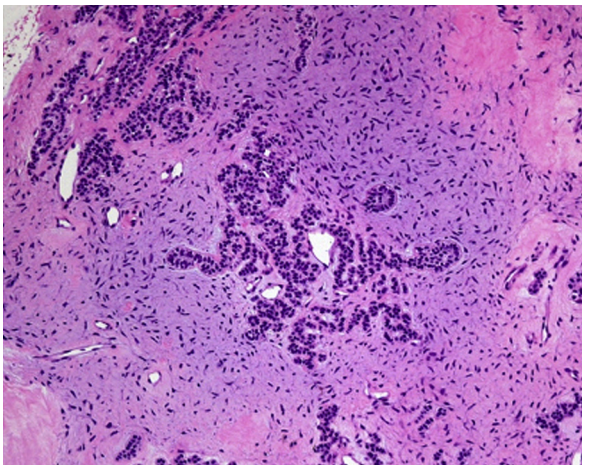

Fig. 4. The tumor consisted of odontogenic ectomesenchymal tissues and odontogenic epithelium. Strands or cords of neoplastic epithelial components were seen in the cellular odontogenic ectomesenchyme (hematoxylin and eosin 10×).

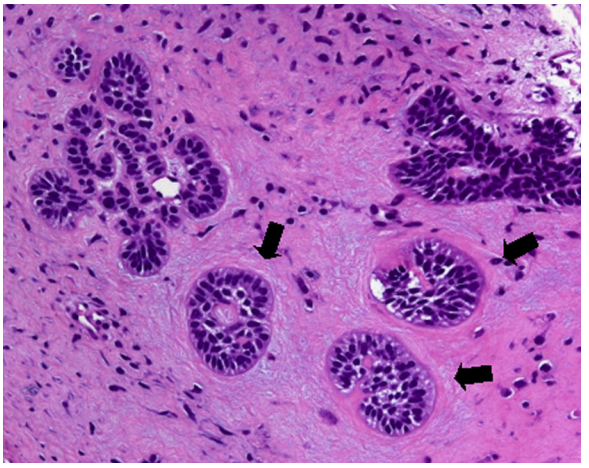

Fig. 5. A peripheral palisading arrangement of the columnar cells was observed in some of the epithelial nests. These resembled an enamel organ and were circumscribed by thin a hyalinous membrane (arrows) (hematoxylin and eosin 20×).

categorized as benign odontogenic mixed tumors. They are composed of odontogenic epithelium and odontogenic ectomesenchyme, with (AFD and AFO) or without (AF) dental hard tissue formation. AFD includes only dentin formation, while AFO possesses the ability to cause induction changes that lead to the formation of both enamel and dentin.[5] In the present case, the tumors were shown to be mixed tumors that consisted of odontogenic epithelium and odontogenic ectomesenchyme. Although formation of dentin-like hard tissue was recognizable, neither the tooth germ-structure nor any induction of enamel occurred in the tumor. Based on these findings, a final diagnosis of AFD was made.