



Contents lists available at ScienceDirect

Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology

journal homepage: www.elsevier.com/locate/jomsmmp

Case Report

A case of a calcifying cystic odontogenic tumor with odontoma in a 5-year-old boy



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ARTICLE INFO

Article history:

Received 31 August 2015
Received in revised form 14 January 2016
Accepted 31 March 2016
Available online 4 May 2016

Keywords:

Calcifying cystic odontogenic tumor
Odontoma
Deciduous dentition

ABSTRACT

We report a case of calcifying cystic odontogenic tumor (CCOT) with odontoma developing in the deciduous dentition period, and present previously reported cases in Japan. A 5-year-old boy visited a dental clinic because of swelling on his left mandibular buccal alveolar region, and was referred to our department. Clinical examination revealed a 20-mm (in diameter), bone-like, hard mass on the left mandibular buccal gingiva at the first deciduous molar region. A panoramic radiograph showed a 25 mm × 20 mm, unilocular, cystic radiolucent area from the left mandibular second deciduous molar to the first molar region. There were radiopaque tiny flecks at the upper side of the lesion. The clinical diagnosis was a benign tumor of the left mandible. Consequently, tumor enucleation was performed under general anesthesia. Histopathological diagnosis confirmed a CCOT associated with odontoma. Five years after surgery, the left mandibular region has healed well, with no evidence of recurrence.

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1. Introduction

Calcifying cystic odontogenic tumor (CCOT) is a disease displaying neoplastic potential and is characterized by calcification of the epithelium and the presence of ghost cells. Although various susceptible ages have been reported, CCOT tends to occur frequently in the young. However, cases of CCOT in patients aged <10 years are rare; in particular, there is only one reported case in Japan of CCOT occurring in the deciduous dentition period [1]. We report our experience of a case of CCOT with odontoma developing in

the deciduous dentition period in a 5-year-old boy, and discuss the relevant literature.

2. Case report

The patient is a 5-year-old boy. In November 2008, he visited a dental clinic to undergo radiography for dental caries. These images indicated the presence of a radiolucent area in the left mandible, and the patient was referred to our department for detailed examination. He did not present with any particular past history or family history of note. He had a height of 100 cm, weight of 15 kg, and low body mass index, but good nutritional status. The external orofacial appearance showed bilateral asymmetry. Regional lymph nodes were without swelling or tenderness. The left mental nerve region did not demonstrate hypoesthesia. The internal oral cavity examination revealed a bone-like, hard mass 20 mm in diameter in the buccal alveolar region of the left mandibular second deciduous molar; however, no parchment-like appearance was observed.

A panoramic radiograph showed a 25 mm × 20 mm, unilocular, cystic radiolucent area from the left mandibular second deciduous molar periapical region to the first molar region. There were radiopaque tiny flecks at the upper side of the area. The

Abbreviation: CCOT, calcifying cystic odontogenic tumor.

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<http://dx.doi.org/10.1016/j.joms.2016.03.009>

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impacted left mandibular second premolar adjacent to the tumor, the mandibular first molar, and the mandibular second molar were deviated. The left mandibular second deciduous molar adjacent to the tumor demonstrated distal root resorption; however, there was no mobility, and a vital reaction was observed (Fig. 1). Computed tomography showed a unilocular, cystic radiolucent area from the left mandibular second deciduous molar periapical region to the distal root, with buccolingual mandibular protrusion, cortical bone resorption, and cortical bone thinning. At the upper side of the cystic radiolucent area, there was a tooth-like radiopaque area. The size of the tumor was 25 mm × 20 mm × 19 mm (Fig. 2). Based on the above findings, we made a clinical diagnosis of a tumor of the left mandible.

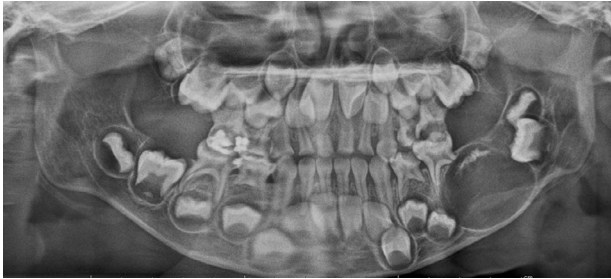


Fig. 1. Panoramic radiograph at the initial examination. A panoramic radiograph showed a 25 mm × 20 mm, unilocular cystic radiolucent area from the left mandibular second deciduous molar to the first molar region. There were radiopaque tiny flecks at the upper side of the lesion.

In December 2008, we performed enucleation of the mandibular tumor under general anesthesia. We also performed a Neumann incision from the medial buccal aspect of the left mandibular second deciduous molar to the alveolar crest, corresponding to the distal buccal aspect of the first molar. When we raised the mucoperiosteal flap, we observed alveolar bone protrusion and partial thinning. Following resection of the buccal alveolar bone, we bluntly detached the tumor from the surrounding bone; we then enucleated the tumor along with the hard tissue inside it en bloc, and closed the wound with absorbable sutures. The fluid in the tumor was yellowish-brown and serous. The tumor was detached easily from the impacted left mandibular second premolar and first molar, which were adjacent to the tumor, and demonstrated partial exposure of the tooth germ in the cavity after enucleation of the tumor, as well as from the distal root of the left mandibular second deciduous molar and the surrounding bone surface, which were adjacent to the tumor. No adhesion was observed. Therefore, rather than removing the left mandibular second premolar, first molar, and second deciduous molar, we chose to retain them. Currently, at five years after surgery, the deviated left mandibular first molar has erupted to the normal location, and the left mandibular second molar has demonstrated movement in a normal direction. In addition, the distal root of the left mandibular second deciduous molar, and the distal root that had demonstrated non-physiological resorption due to the lesion were being lost following the progression of physiological resorption; the permanent left second premolar has erupted to the normal location. We have not performed occlusal guidance for the left mandibular second premolar, first molar, or second molar. The patient is now 10 years old, with no apparent recurrence, and has made favorable progress (Fig. 3). The resected material comprised hard tissue demonstrating a cystic structure 20 mm × 20 mm in size; in the lumen of the cyst wall-like structure, hard tissue with an irregular surface was observed (Fig. 4). The hard tissue, in which a cyst wall-like structure was observed, was histologically an odontoma composed of dentin with a tubular structure, an enamel matrix structure, and a cementum

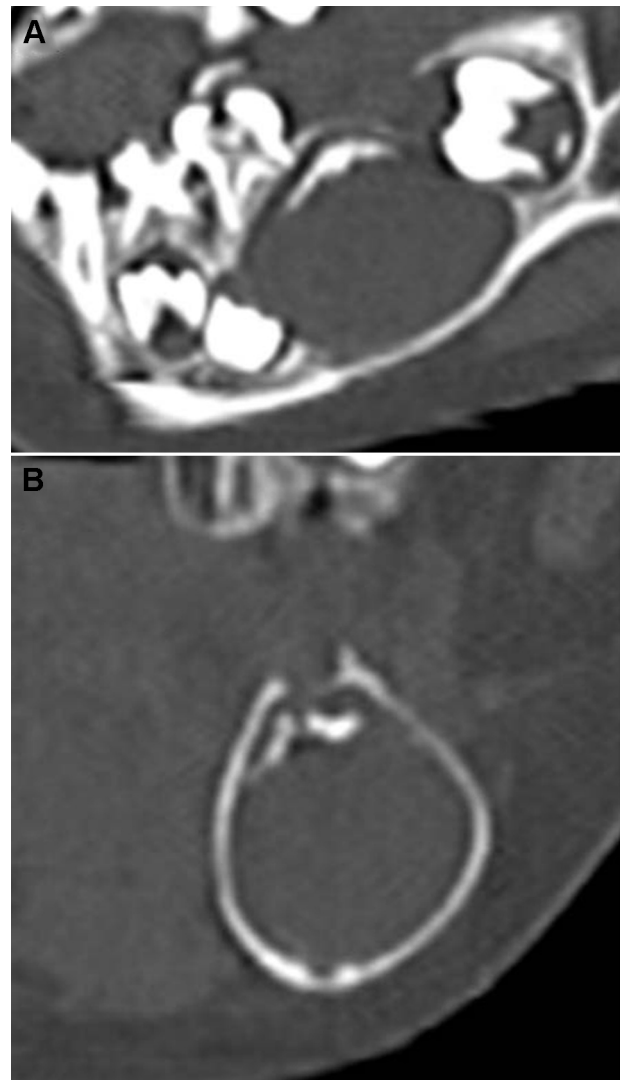


Fig. 2. CT images. (A) Sagittal image. A unilocular cystic radiolucent area was observed from the left mandibular second deciduous molar periapical region to the distal root. At the upper side of this area, there was a tooth-like radiopaque area. (B) Axial image. Buccolingual jawbone protrusion, cortical bone resorption, and cortical bone thinning occurred. The mandibular canal is compressed and deviated at the inferior border of the mandible by the tumor.

structure. The cyst wall-like tissue encapsulating this odontoma demonstrated an odontogenic proliferative epithelium lining the inner surface. The lining epithelium, composed of cuboidal and stellate reticulum cells, contained ghost cells, which were eosinophilic and swollen. The nuclei of the ghost cells disappeared, leaving



Fig. 3. Panoramic radiograph 5 years after surgery. The tumor cavity disappeared; the left mandibular second premolar and first molar, which had deviated due to the tumor, erupted to their normal locations; the left mandibular second molar moved in a normal direction.

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