

Compound odontoma in the primary dentition: A case report of a 4-year-old girl

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Odontomas have been classified as odontogenic benign tumors composed of dental tissue. These tumors are generally asymptomatic and seen most commonly in association with unerupted teeth. In general, odontomas occur more often in the permanent dentition, and they are rarely associated with the primary dentition. The present report describes the unusual occurrence of a compound odontoma in a 4-year-old female who had an unerupted mandibular left primary canine and painless swelling in that region noticed by parents. Her clinical examination demonstrated a well-defined painless swelling with normal overlying mucosa, firm in consistency, extending from the primary lateral incisor to the primary first molar at the same site with buccal expansion, normal temperature, and non-pulsatile. Radiographic examination revealed the presence of multiple radiopaque tooth-like structures near the crown of the primary unerupted canine. Surgical removal of the tumor was performed under general anesthesia and the histological examination confirmed the diagnosis of a compound odontoma.

Key words: Compound odontoma, primary dentition, unerupted primary tooth

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Introduction

Odontomas are the most prevalent (~36–75%) type of odontogenic tumor [1]. Odontomas are defined as mixed tumors composed of odontogenic epithelium and mesenchymal tissue [2]. The etiology of odontomas is still unclear; however, factors such as trauma, local infection, genetic mutation, and hereditary can lead to lesion formation [3]. The World Health Organization has subdivided odontomas into compound and complex types based on histologic criteria [4]. Compound odontomas are similar to normal teeth, while complex odontomas are irregular masses [5]. These tumors

are mostly associated with the permanent teeth and are rarely associated with primary teeth [4-6]. Impacted teeth and delayed eruption due to an odontoma in the primary dentition is unusual [7-10]. There is no sex-based predilection for odontoma development [11]. Early diagnosis of odontomas in the primary dentition is essential to prevent complications, such as impaction or failure of the primary or permanent teeth to erupt [12]. The post-treatment prognosis is very favorable and the recurrence rate is extremely low [13-14]. In this case report, the diagnosis and management of a compound odontoma, which is very rarely diagnosed associated with unerupted primary mandibular canine, is presented.

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Case report

A girl aged 4 years 3 months presented to the Pediatric Dental Clinic of the Faculty of Dentistry, Mahidol University with an unerupted mandibular left primary canine and a painless swelling in that region that had been noticed by her parents when the patient was 6 months old. Her medical history was unremarkable. There was no history of trauma to her oro-facial region. There was no family history of a similar condition. This was her first dental visit. On intra-oral examination, multiple carious anterior upper teeth with a missing mandibular left primary canine and an ~2 mm midline shift of the mandibular arch to the left side. A 1×1 cm well-defined painless swelling with normal overlying mucosa, which was firm in consistency, extended from the primary lateral incisor to the primary first molar at the same site with buccal expansion, normal temperature and non-pulsatile (Figure 1). Periapical and panoramic radiographic examination revealed the presence of multiple radiopaque tooth-like structures near the crown of the primary unerupted

canine (Figure 2). Our differential diagnosis based on the clinical and radiographic findings was a compound odontoma. The patient was scheduled for surgical removal of the lesion.

The surgical removal was performed under general anesthesia. A buccal mucoperiosteal flap was raised in the lower left canine region. Bone was removed with a bur and multiple odontomas and the mandibular left primary canine were removed (Figures 3, 4). The surgical wound was closed primarily with 4/0 vicryl sutures. The lesion consisted of several tooth-like structures and was sent for histological assessment. The histopathological examination



Figure 1 Intraoral view showing the unerupted left lower canine with a swelling in that region.

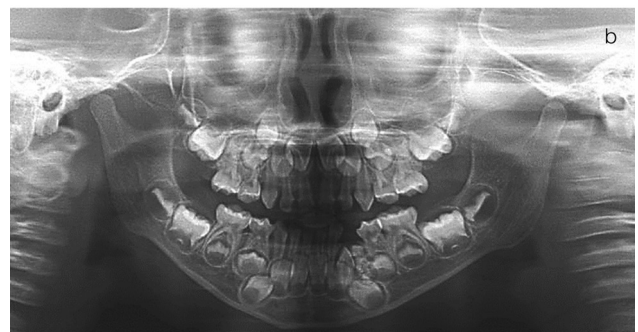


Figure 2 (a,b). Periapical and panoramic radiographs revealed the presence of multiple radiopaque tooth-like structures near the crown of the unerupted mandibular primary canine.

revealed that the specimen consisted of dentin masses, developing crowns of tooth-like structures in normal and abnormal relationships and cementum-like masses and ghost cells, with occasional squamous epithelial cells (Figure 5). The histological findings confirmed the diagnosis of a compound odontoma.

At the 2-year follow-up after the odontoma was removed, panoramic radiographic examination revealed no signs of recurrence (Figure 6). In addition, there was inadequate space for the permanent canine to erupt. Further malocclusion might be corrected by orthodontic treatment.

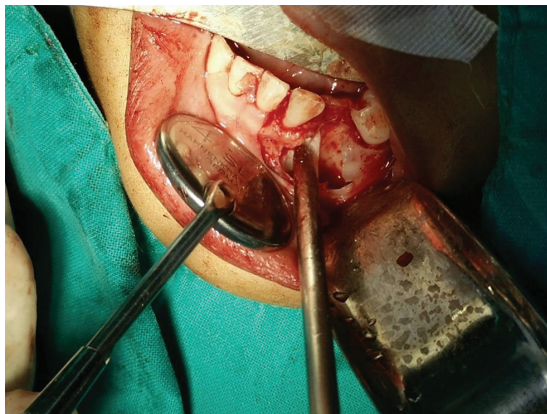


Figure 3 A buccal mucoperiosteal flap was raised.



Figure 4 A compound odontoma was removed after elevating the mucoperiosteal flap.

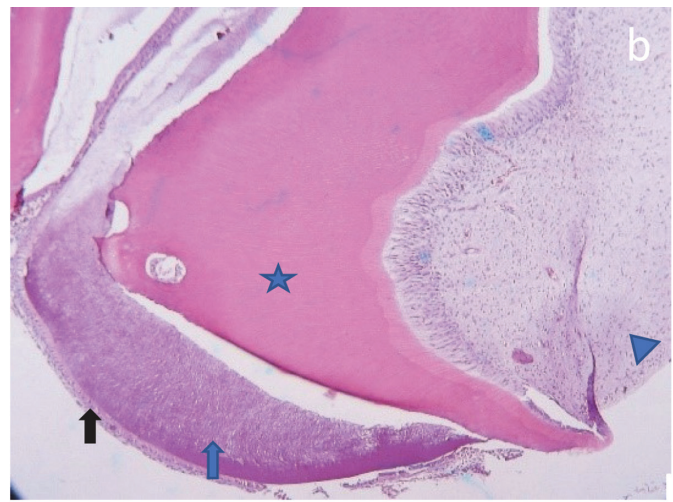
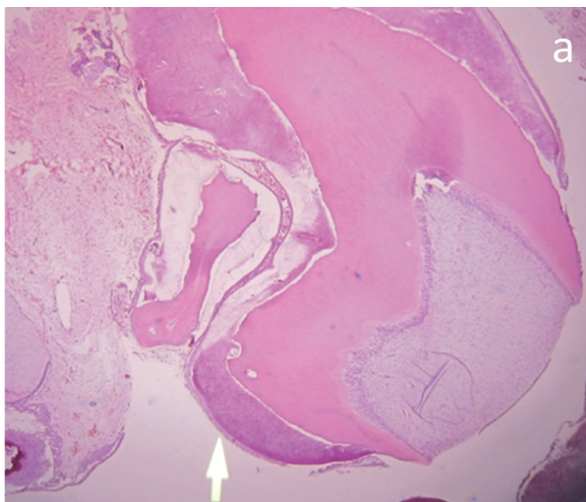


Figure 5 a: H&E×4 magnification, tooth-like structures (white arrow), b: H&E×10 magnification, follicular epithelium (black arrow), enamel matrix (blue arrow), mineralized dentin (star), dental pulp (arrowhead). The specimen consisted of dentin masses, developing crowns of tooth-like structures in normal and abnormal relationships. Cementum-like masses and ghost cells, with squamous epithelial cells are also noted occasionally.



Figure 6 Panoramic radiograph revealed no signs of recurrence at the 2-year follow-up after treatment.

Discussion

Odontomas are the most common type of odontogenic tumors. Odontomas frequently lead to impaction or delayed eruption of the permanent teeth; however, this rarely occurs in the primary dentition as observed in this case [4-6]. Compound odontomas are equally distributed between males and females [11]. However, some studies reported a higher prevalence in females, while others reported a higher prevalence in males [1]. These disparate findings are likely due to racial/ethnic differences between study populations.

Most studies have found that odontomas occur more often in the maxillary arch; however, some studies reported no differences between the two jaws [14]. In our case, the odontoma was located in the mandibular arch. Compound odontomas most commonly occur in the incisor-canine region [15], which was also seen in this case. However, the tendency of odontomas to occur in the right side of jaw was in contrast to our finding [3]. Odontomas are generally asymptomatic [5] as observed in this case.

The radiographic differential diagnosis of a compound odontomas is very limited because they appear as radiopaque masses as collection of small tooth-like structures, however, with a complex odontoma a broader differential diagnosis should be considered because they appear as a unique radio-opacity [1]. In this case, the radiographic examination revealed the presence of multiple radiopaque tooth-like structures near the crown of the primary unerupted canine.

The differential diagnosis should include ameloblastic fibroma, ameloblastic fibroodontoma, and odontoameloblastoma, which may occur in association with syndromes, such as Gardner syndrome, Hermann syndrome, and basal cell nevus syndrome [16]. In the present case, there was no associated syndrome or history of trauma to the patient. Based on the clinical and radiographic findings, a provisional diagnosis of odontoma was established.

Histopathology analysis of the specimen is helpful to establish the definitive diagnosis for odontoma lesion [12]. Here, the diagnosis was confirmed by an oral pathologist as a compound odontoma.

The treatment of choice for a compound odontomas is surgical removal, followed by histopathological analysis to confirm the diagnosis [17]. Based on our literature review, the optimal management of an unerupted tooth is to allow it to remain and reposition itself in the arch [17]. In contrast, unerupted teeth are frequently reported to be extracted simultaneously with the odontoma [18]. In this case, the primary canine was mobile while removing the odontoma. This made the tooth difficult to preserve and therefore it was extracted. Space management might be needed by this patient in the future.

The prognosis after odontoma removal is very favorable and the recurrence rate is extremely low [18]. Indeed, this patient's panoramic radiographic examination revealed no signs of recurrence at the 2-year follow-up after treatment.

Odontomas occur mainly in the permanent dentition with very few reports mentioning the occurrence of this lesion in the primary dentition. In our case, the cause of the unerupted primary canine was a compound odontoma. For early detection of odontomas, radiographic and careful clinical examinations are important. Complications can be prevented by surgical excision followed by histopathological analysis. A careful follow-up of the case, both clinically and radiographically is necessary to assess prognosis.

Conclusion

Early diagnosis and treatment of compound odontomas in the primary dentition are necessary to prevent any later complications, such as failure of eruption of primary and permanent teeth.

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