Odontomas in the Primary Dentition: Literature Review and Case Report

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ABSTRACT

Odontomas are hamartomatous developmental malformations of dental tissues. They are very rarely diagnosed in the primary dentition. The purpose of this paper was to review the literature in relation to odontomas in the primary dentition, and describe the dental management of a child, age 4 years and 8 months, who had a complex odontoma. (*J Dent Child*. 2004;71:73-76)

KEYWORDS: COMPLEX ODONTOMA, PRIMARY DENTITION

Odontomas are hamartomatous developmental malformations of dental tissues and not neoplasms.[1] They arise from the epithelial and ectomesenchymal odontogenic tissues. Odontomas usually develop and mature at the same time as their adjacent teeth. Therefore, they must arise from dental lamina or ectopic residues of odontogenic epithelium at the same time as adjacent teeth. The lesions are known as compound odontomas if the dental tissues are deposited in the form of small tooth-like structures or denticles. If the hard and soft dental tissues are distributed irregularly throughout, the lesions are termed complex odontomas. Both may be associated with unerupted permanent teeth, particularly, canines, maxillary central incisors, and third molars. Odontomas may also be associated with failure of eruption of primary teeth,[2] missing permanent teeth, dentigerous cyst formation, and rarely calcifying odontogenic cysts.[3] They are generally asymptomatic, but occasionally give rise to pain and swelling.[4] The etiology of odontomas is unknown, but genetic factors and environmental causes, such as trauma and infection, have been proposed. The purpose of this paper was to review the literature in relation to odontomas in the primary dentition and describe the dental management of a child, age 4 years and 8 months, who presented with a complex odontoma.

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LITERATURE REVIEW

Odontomas are usually evident in the second decade of life, but may be discovered at any age. They are frequently located in the anterior of the maxilla or the posterior of the mandible. Odontomas are very rarely diagnosed in the primary dentition. A summary of cases diagnosed in the primary dentition is shown in Table 1. Eleven of the lesions were present in males and 6 in females. The majority were compound odontomas and located in the maxillary primary canine region, and, less frequently, in the incisor and molar areas. Of the 17 cases reviewed here, 14 were associated with failure of eruption of a primary tooth, 2 with a missing primary tooth, 1 with a calcifying odontogenic cyst, and only 1 with asymmetrical spacing in the dental arch. The patients were managed by enucleation of the odontomas, and in only 4 cases were the associated underlying primary teeth extracted.[3,6,7,9]

CASE REPORT

A female, age 4 years and 8 months, was referred by a specialist in pediatric dentistry to the Department of Pediatric Dentistry, Guy's Hospital, for management of dental caries and an ill-defined radiolucent lesion in the maxillary right primary canine region. Her mother was white and her father was Asian. Both parents were concerned about their daughter's "missing tooth" in the space distal to the maxillary right primary canine. The patient's medical history was normal.

Extra-oral examination revealed right submandibular lymphadenopathy. Intraorally, all the primary teeth were present and there were caries in the primary molars, some of which had been restored temporarily. There was a space of approximately 6 mm in the maxilla between the distal surface of the right primary canine and the mesial surface of the right first primary

Table 1. Summary of Case Reports of Odontomas in the Primary Dentition

Publication	Age and gender of patients*	Type of odontoma	Location and presentation	Management and outcome
Axel AL, 1937[5]	4-year-old male*	Compound ("supernumerary teeth in a cyst")	Located in the region of a missing maxillary left primary canine.	Surgical removal of the lesion. No follow-up of the case was mentioned.
Aimes ABP, 1947[6]	4-year-old male*	Compound	The maxillary right primary lateral incisor to the maxillary primary molar region. Failure of eruption of the maxillary right primary canine.	Surgical removal of the lesion, the maxillary right primary molars, and the primary canine.
Aimes ABP, 1952[7]	3-year, 6 month-old male*	Compound	The maxillary right primary central incisor to the second primary molar region.	Surgical removal of the lesion, the maxillary right primary incisors, and canine.
Hitchin AD, White JW, 1955[8]	4-year-old male*	"Dentinoma" (Complex)	Mandibular left primary lateral incisor causing failure of eruption of this tooth.	Surgical removal of the odontoma. No follow-up mentioned in relation to eruption of the primary lateral incisor.
Hitchin AD, Dekonor E, 1963[9] (2 cases)	4 year, 11 month-old male*	Compound	Maxillary left primary canine region with failure of eruption of this tooth.	Surgical removal of the lesion. No follow-up mentioned in relation to eruption of the primary canine.
	8 year, 7 month-old female*	Compound	Lesion first noticed by parents when patient was 5 years of age when there was failure of eruption of the maxillary right primary canine.	Surgical removal of the odontoma and the maxillary right primary canine. No follow-up mentioned in relation to eruption of the maxillary permanent canine.
Noonan RG, 1971[10]	5-year-old Afro-Caribbean female	Compound	Maxillary right primary canine region causing failure of eruption of the primary canine.	Surgical removal of the odontoma resulted in spontaneous eruption of the primary canine 3 weeks later.
Levine N, Stoneman DW, 1977[11]	2 1/2-year-old White male	Compound	Maxillary right primary lateral incisor region causing failure of eruption of this tooth.	Surgical removal of the odontoma, after which the primary lateral incisor erupted.
Stajcic ZZ, 1988[12]	6-year-old male*	Compound	Located in the region of a missing maxillary left primary lateral incisor.	Surgical removal of the odontoma.
Brunetto AR et al, 1991[13]	2-year old White male	Not stated	Maxillary right primary canine region causing failure of eruption of tooth.	Surgical removal of the odontoma and orthodontic extrusion of the primary canine.
Haishima K et al, 1994 (2 cases)[14]	1 year, 2 month old Japanese female	Compound	The maxillary left primary central incisor region and failure of eruption of this tooth in both cases.	Surgical removal of the odontomas. Primary central incisors erupted spontaneously, but only following surgical exposure of the incisor 9 months after removal of the odontoma in the older patient.
	1 year, 8 month old Japanese male	Compound		
Bacetti T, 1995[15]	3 year, 6 month old female*	Compound	Maxillary right primary canine region causing failure of eruption of the primary canine.	Surgical removal of the odontoma resulted in spontaneous eruption of the primary canine after 2 years.
Olivero JAGP et al, 1995[3]	3-year-old female*	Compound odontoma with a calcifying odontogenic cyst	Failure of eruption of a maxillary right primary lateral incisor.	Extraction of the unerupted maxillary right primary lateral incisor, osteotomy, and curettage of the lesion. No recurrence of the lesion after 4 years.
Long WR et al, 1998[16]	30 month old White male	Compound	Maxillary left primary molar region resulting in spacing between the first and second primary molars.	Surgical removal of the lesion.
Motokawa W et al, 1998[17]	3-year-old Japanese female	Complex	Maxillary left second primary molar causing failure of eruption of this tooth.	Surgical removal of the odontoma. Eight months later the maxillary left second primary molar, which had failed to erupt, was surgically exposed and orthodontically repositioned.
Yassin OM, 1999[18]	4-year-old Middle-Eastern male	Compound	Maxillary right primary canine region causing failure of eruption of the primary canine.	Surgical removal of the odontoma resulted in spontaneous eruption of the primary canine.

*ethnicity of patients was not always stated by the authors

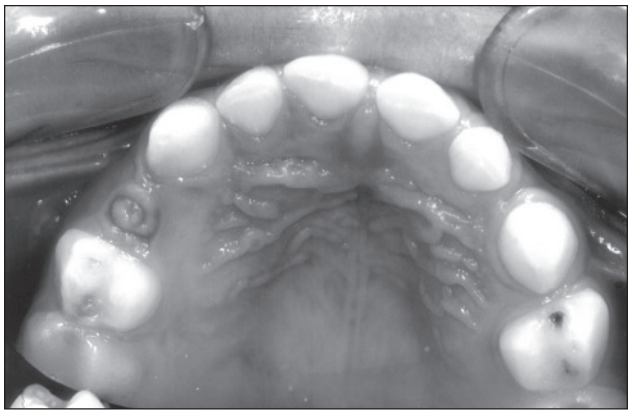

Figure 1. Unilateral spacing is seen in the maxilla between the right primary canine and right primary first molar. The overlying mucosa is raised and inflamed.

molar. A similar space was absent on the contralateral side of the arch. Soft tissue examination revealed a non-draining soft tissue swelling of approximately 3 mm in diameter on the alveolar ridge in this region. The overlying mucosa was red and inflamed, but the lesion was symptomless (Figure 1).

A dental panoramic tomograph showed the presence of all primary and permanent teeth, except the third molars. Posterior bite-wing radiographs revealed an irregular radio-opaque mass of approximately 6×5 mm distal to the maxillary right primary canine and confirmed the presence of dental caries in the primary molars (Figure 2). A provisional diagnosis of an odontoma was made, and the patient was scheduled for enucleation of the lesion and oral rehabilitation under general anesthesia.

The primary molars were restored with stainless steel crowns, amalgam and preventive resin restorations. A mucoperiosteal flap was elevated in the maxillary right primary canine and first primary molar region and a calcified mass removed (Figure 3). The flap was replaced and sutured with 2 3/0 Vicryl sutures. The child's recovery was normal and intraoral healing was satisfactory at a 3-month follow-up visit. The patient was subsequently referred back to her own dentist for prevention and continuing care.

HISTOLOGICAL EXAMINATION

Macroscopic examination revealed an irregular yellow hard tissue mass (9×7×5 mm) with pale soft tissue at one end (Figure 4). Microscopic examination revealed an irregular mass

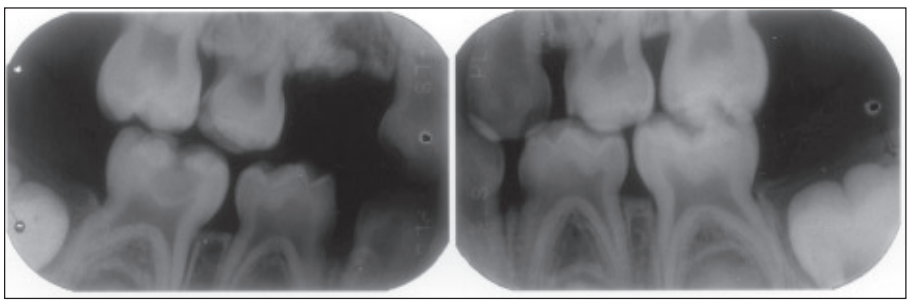

Figure 2. Right and left posterior bite-wing radiographs show an irregular radio-opaque mass distal to the maxillary right primary canine and caries in the primary molars.

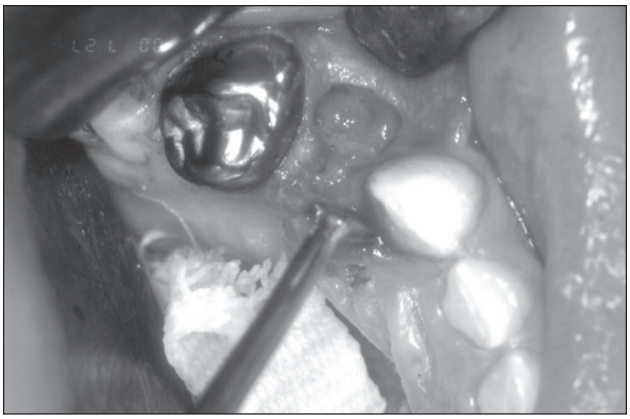

Figure 3. Enucleation of the odontoma.

of dysplastic dentin containing spaces filled with enamel matrix and partially reduced enamel epithelium (Figure 5). A diagnosis of a complex odontoma was made.

DISCUSSION

Odontomas were reported to be the most common odontogenic tumors in North America.[19, 20] Diagnosis of odontomas in the primary dentition, as in the present case, is unusual. A retrospective study was conducted of 250 oral and maxillofacial tumors in children, ages 4 months to 15 years, who attended the Tokyo Medical & Dental University between 1965 and 1992.[21] Seventy-nine (32%) were odontogenic tumors, of which 47 (19%) were odontomas. The majority of odontomas were seen in 6 to 15 year olds, with only 3 reported in 1 to 5 year olds. In another study of oral and maxillofacial biopsy specimens from a pediatric population in southern Taiwan, 73% of odontomas (29/40) were found in children ages 11 to 15 years, 23% (9/40) in the 6- to 10-year age group and only 5% (2/40) in children ages 3 months to 5 years.[22]

In the present case, the marked asymmetry of the maxillary arch in the region of the unerupted odontoma was somewhat unusual. The authors were aware of only 1 other case with a similar presentation. A 3-year old white male presented with an unerupted compound odontoma between the maxillary left first and second primary molars resulting in arch asymmetry.[16] The lesion was symptomless, and unilateral spacing in the dental arch, in an area not normally expected, led to the diagnosis as in the present case. Both cases were similar in that there was no interference of eruption of the primary teeth, and both were successfully managed by enucleation.

Histological examination of the present case revealed a typical complex odontoma, but the majority of previously reported cases in the primary dentition were compound odontomas. However, the distinction between the 2 types is somewhat artificial, and, in the permanent dentition, many have an intermediate pattern.[1]

This case highlighted the importance of considering the presence of

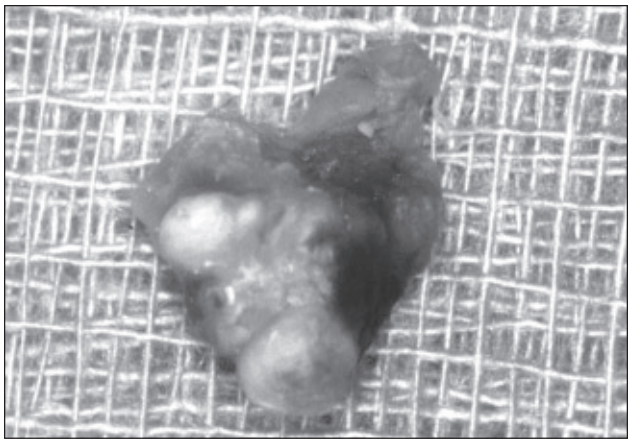

Figure 4. Macroscopic view of the odontoma shows an irregular hard tissue mass with pale soft tissue at one end.

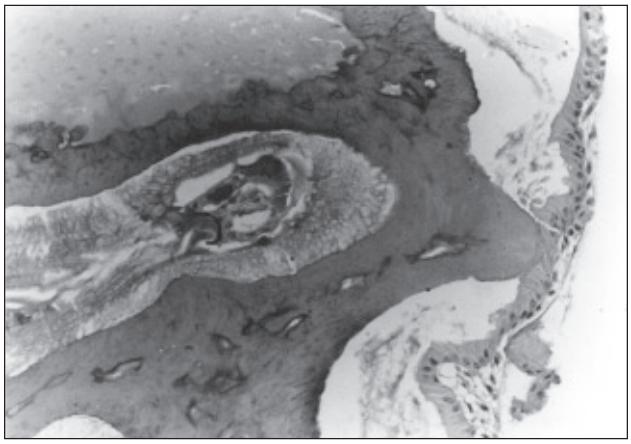

Figure 5. Microscopic view of part of the odontoma shows a disorganized mass of dysplastic dentine, which is sparsely tubular and contains several voids. Centrally within the dentine there is enamel matrix showing normal prismatic structure and a layer of ameloblasts along the surface at the top.

an odontoma as a possible cause of swelling and spacing, giving rise to arch asymmetry in the primary dentition. It also emphasized the importance of radiographs for the diagnosis of hard tissue lesions, including caries, in the primary dentition. Early diagnosis and management of odontomas in the primary dentition are essential in order to prevent later complications, such as failure of eruption of the primary and permanent teeth and potential space loss.

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