



# Varying Presentation of Odontoma in Early Childhood Involving Deciduous Dentition

Himani Kaushik<sup>1</sup> Ruchi Singhal<sup>1</sup> Ritu Namdev<sup>1</sup> Anisha Kakran<sup>1</sup> Shefali Negi<sup>1</sup>

<sup>1</sup>Department of Pedodontics, Post Graduate Institute of Dental Sciences, Rohtak, India

**Address for correspondence:** Himani Kaushik, MDS, Department of Pedodontics, Post Graduate Institute of Dental Sciences, Rohtak, Haryana 124001, India

**E-mail:** shininghemnikaushik@gmail.com

## Abstract

Most common tumors of odontogenic origin are odontomas, which have slow-growing and non-aggressive behavior. In general, odontomas are more commonly associated with mixed and permanent dentition, with very few case reports of them being associated with deciduous dentition. Odontomas should be surgically managed once detected to avoid eruption disturbances and malocclusion. This article presents two case reports of odontomas in early childhood involving primary dentition.

**Keywords:** Deciduous dentition, denticles, early childhood, odontoma, oral lesions, oral pathology

## Introduction

Among the odontogenic tumors, odontomas are the most common benign mixed tumors. Rather than a neoplasm, these are hamartomatous anomalies in which differentiation of the epithelial cells and mesenchymal cells has taken place completely into functional ameloblasts and odontoblasts to form enamel, dentin, and cementum, i.e., histo-differentiation is completed, but the odontogenic cells fail to reach the complete morpho-differentiation.[1] So, enamel and dentin are formed in an abnormal and disorganized pattern. It consists of diverse types of tissues, hence, also called composite odontoma. Odontomas are classified based on alteration and organization of odontogenic cells into compound and complex odontomas. Compound odontomas show histological as well morphological differentiation of the odontogenic cells. Enamel, dentin, cementum, and pulp are well organized and surrounded by follicular connective tissue resembling a tooth-like structure. Complex

odontomas, on the other hand, show histological differentiation only. Enamel, dentin, cementum, and pulp are laid in a disorganized manner as a hard mass not resembling tooth structure.[2] In early stages of the development of odontoma, variable amounts of proliferating odontogenic epithelium and mesenchyme are present, and there is some evidence of calcification. It is seen as a circumscribed radiolucent tumor. Histopathology shows proliferating mesenchyme and odontogenic epithelium, resembling tooth germs.[1]

While there is a dilemma over the etiology of odontomas, they are known to occur due to local trauma, inflammation, infection, hereditary conditions like Hermann's syndrome and Gardner syndrome, hyperactivity of odontoblasts, and mutations in the genes controlling the development of the dentition.[3] It can cause disruption in the normal eruption and shedding of deciduous dentition leading to impacted teeth, delayed eruption, and prolonged retention.[4]

**How to cite this article:** Kaushik H, Singhal R, Namdev R, Kakran A, Negi S. Varying Presentation of Odontoma in Early Childhood Involving Deciduous Dentition. J Pediatr Dent 2024;10(1):50-53

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Supra-eruption of contralateral teeth and inclination of adjacent teeth in the missing space can follow, with primary teeth being impacted due to odontoma.

In general, odontomas are more commonly associated with mixed and permanent dentition, with very few case reports of them being associated with deciduous dentition (Table 1). This article presents a case series of rarely diagnosed odontomas related to primary dentition.

### **Case 1: Compound Odontoma Associated with an Unerupted Primary Maxillary Canine**

A girl, four years of age, reported with a chief complaint of swelling in the upper right jaw region for the last 5 months. There was no relevant history of trauma or infection and no associated family history. Extra-orally, the face was found symmetrical. Intra-oral examination revealed a well-defined swelling of around 1.5 cm diameter in the right maxillary vestibular region in relation to clinically missing right primary maxillary canine with normal appearing overlying mucosa (Fig. 1a). The swelling was round, non-tender, firm, and non-compressible. Radiographic examination showed multiple radio-opaque tooth-like masses around the crown of the unerupted primary upper canine (Fig. 1b). Compound odontoma was the provisional diagnosis based on the above findings. Surgical removal of the tooth-like structure was planned under local anesthesia. After raising the mucoperiosteal flap, multiple odontomas were removed with a periosteal elevator (Fig. 1c). The associated primary canine was not extracted during the surgery to explore the possibility of its spontaneous physiological eruption in the future. The surgical wound was closed using 3-0 silk sutures. After 7 days, sutures were removed. Healing was reported to be uneventful. The patient was kept on regular follow-up. At 6-months follow-up, the primary canine was seen to be erupting in the oral cavity (Fig. 1d). After 1 year, the canine was fully erupted in the dental arch (Fig. 1e).

### **Case 2: Developing Odontoma in Mandibular Anterior Region**

A female patient, 3 years and 5 months of age, reported with a chief complaint of swelling in the left lower teeth region since 2 months, which was painful when touched. On extra-oral examination, the face was apparently symmetrical. Intraoral examination showed a round swelling of around 2.5 cm present in relation to the left mandibular lateral incisor, canine, and first molar, obliterating the vestibule with overlying mucosa reddish-colored, which was firm on palpation, nontender, non-reducible,

non-compressible swelling (Fig. 2a). All the primary teeth in the left lower quadrant were caries-free and firm in their sockets. Parents were advised for radiographic examination but could not be done successfully due to the non-cooperation of the patient. The provisional diagnosis of a benign lesion of dental hard tissues with cystic changes was made, and surgical enucleation of the lesion under general anesthesia was planned (Fig. 2b-d). Tissue obtained on enucleation was sent for histopathological examination (Fig. 2e). Histopathological report revealed well-arranged odontoblastic layer with cells exhibiting reverse polarity and a thin layer of predentin at the junction between connective tissue and odontoblastic layer. Thin sheets of dentin were laid in different proportions at various sites. One focus showed predentin lying superficially over the connective tissue. Other foci showed ectomesenchyme along with a few hyperchromatic cuboidal epithelial cells mimicking cells of odontogenic origin. Inflammatory cystic lining was also evident. Cystic changes disturbed the maturation of the developing odontoma (Fig. 2f, g). Diagnosis of developing odontoma with tooth-like calcification and haphazardly formed dentin was made. On follow-up after a year, a radiograph depicted the absence of permanent tooth germ in relation to the left primary first molar, which may be caused by degeneration of the tooth bud by developing odontoma or idiopathic absence of permanent tooth or removal of the crypt of premolar during surgical enucleation.

### **Discussion**

The majority of odontogenic tumors are odontomas (22%), mostly diagnosed in the second decade of life at a mean age of 14 years.[2,5,6] Both the cases discussed here were seen in primary dentition. In case 1, a compound odontoma was diagnosed at the age of 4 years, whereas in Case 2, a developing odontoma was found at the age of 3.5 years.

The most common location for the occurrence of odontoma is the upper jaw in the incisor-canine area, followed by the anterior region in the lower jaw.[1,2] It may disturb the eruption of teeth leading to delayed eruption, impaction, or over-retention of deciduous teeth. In the first case, odontoma was found to be the cause of impaction of the maxillary deciduous canine.

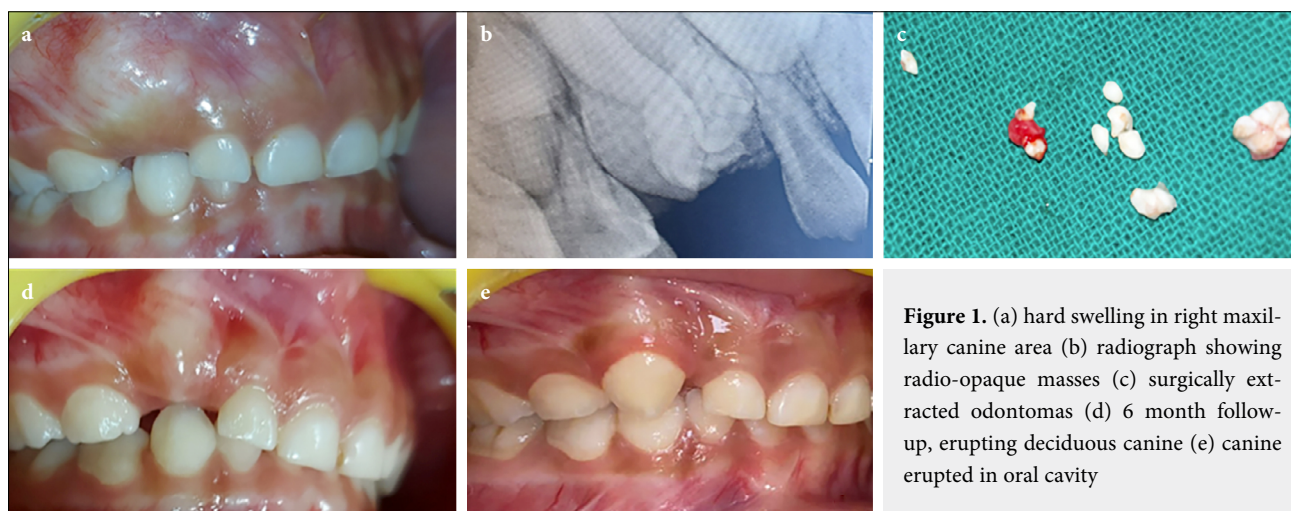
Based on the degree of calcification, the development of odontoma can be described under three stages. The first stage of the lesion is characterized by a radiolucent appearance due to a lack of calcification. Partial calcification of the lesion is seen in the intermediate stage, resembling

**Table 1.** Cases of odontoma associated with primary dentition described in literature

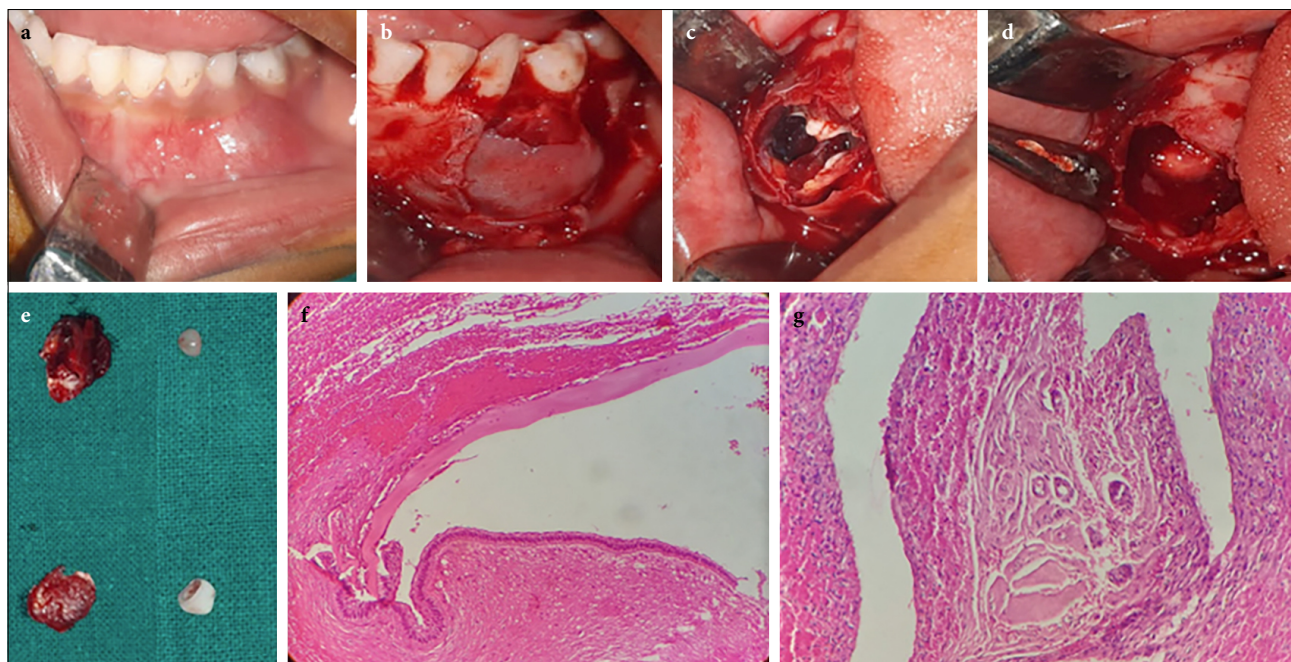
Author	Age of patient	Location	Type
Axel (1937)	4-years	Maxilla	Compound odontoma
Aimes (1947)	4-years	Maxilla	Compound odontoma
Aimes (1952)	3-years, 6months	Maxilla	Compound odontoma
Hitchin - White (1955)	4-years	Mandible	Complex odontoma
Hitchin-Dekonor (1963)	4-years, 11-months	Maxilla	Compound odontoma
Hitchin-Dekonor (1994)	8-years, 7-months	Maxilla	Compound odontoma
Noonan (1971)	5-years	Maxilla	Compound odontoma
Stajcic (1988)	6-years	Maxilla	Compound odontoma
Brunetto (1991)	2-years	Maxilla	Compound odontoma
Haishima (1994)	1-year, 2-months	Maxilla	Not mentioned
Haishima (1994)	1-year, 8-months	Maxilla	Compound odontoma
Bacetti (1995)	3-years, 6-months	Maxilla	Compound odontoma
Olivero (1995)	3-years	Maxilla	Compound odontoma
Long (1998)	30-months	Maxilla	Compound odontoma
Motokawa (1998)	3-years	Maxilla	Complex odontoma
Yassin (1999)	4-years	Maxilla	Compound odontoma
Yeung (2003)	2-years, 5-months	Maxilla	Compound odontoma
Sheehy (2004)	4-years, 5-months	Maxilla	Complex odontoma
Nelson-Filho (2005)	1-year, 8-months	Maxilla	Compound odontoma
Yildirim (2007)	4-years	Mandible	Compound odontoma
Teruhisa (2009)	6-years	Mandible	Compound odontoma
Singla (2017)	6-years	Maxilla	Compound odontoma
Salim (2021)	5-years	maxilla	Compound odontoma
Salim (2021)	4-years	maxilla	Compound odontoma
Ahammed (2021)	5-years	Mandible	Complex odontoma

calcifying cystic odontogenic tumors, fibro-osseous lesion, chronic osteomyelitis, and fibrous dysplasia. Odontoma appears radio-opaque in final stages with a radiolucent halo surrounding it. These tumors are shown to be

related to other odontogenic cysts and tumors such as ameloblastoma, ameloblastic fibro-odontoma, odontoameloblastoma, calcifying epithelial odontogenic tumor, adenomatoid odontogenic tumor, and dentigerous cyst.[7]



**Figure 1.** (a) hard swelling in right maxillary canine area (b) radiograph showing radio-opaque masses (c) surgically extracted odontomas (d) 6 month follow-up, erupting deciduous canine (e) canine erupted in oral cavity



**Figure 2.** (a) hard swelling in left mandibular primary canine-First molar area (b-d) during surgery (e) surgically excised mass with 2 denticle (f, g) odontogenic epithelial and dentinoid like material in the form of developing odontome and underlying haemorrhagic stroma

WHO omitted ameloblastic fibrodentinoma, ameloblastic fibro-odontoma, and odontoameloblastoma from its latest classification (2017) because of their overlapping histological features and precursors with mature odontoma.[8]

Due to interactions between epithelium and ectomesenchyme in the odontogenic tumors, there can be the formation of dental tissues to varying extents, as seen in Case 2. The epithelium in odontogenic tumors has the tendency to proliferate until the completion of odontogenesis (up to the age of 22 years),[9] so early removal and thorough curettage of the epithelium should be done to check for any signs of recurrence on the follow-up visits.

Early diagnosis and early removal of the odontoma are essential to improve the prognosis of the involved teeth. Early treatment allows the impacted tooth to restart the physiological eruption, as seen in the first case. When there is no sign of eruption spontaneously, orthodontic forces should be used for management. Extraction of the tooth should be the last resort.

## Conclusion

Early detection and management of odontoma should be done to avoid complications related to the teeth involved, subsequent malocclusion, and arising pathological complications. As the tumor can be present in deciduous dentition, dentists and parents should be aware of the lesion for early management.

**Financial Disclosure:** Nil.

**Conflict of Interest:** None declared.

**Use of AI for Writing Assistance:** Not declared.

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