An Impacted Malformed Primary Maxillary Central Incisor Diagnosed as a Compound Odontoma.

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An odontoma is usually found in the surrounding area of the unerupted permanent tooth bud and rarely adjacent to the primary central incisor. This report presents a case of an unerupted malformed primary central incisor, histologically diagnosed as a compound odontoma. A 5.5 year-old healthy boy presented with an unerupted maxillary left primary central incisor and mild gingival buccal swelling. No history of traumatic injury was recorded. Radiological examination revealed a tooth-like mass with a partially developed root and a malformed crown in the area of the left primary central incisor. Delayed development of the successor left permanent incisor in comparison to the right one was noted. The rational for early surgical intervention to enable normal development and eruption of permanent incisors is described. Clinical and radiographic diagnoses as well as several etiological factors and a differential diagnosis are considered and reviewed.

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INTRODUCTION

he normal eruption pattern of upper primary central incisors occurs around the age of 6-8 months, usually when two thirds of the root has already developed. However, variations in the eruption sequence and appearance of teeth in the oral cavity are common.^{1,2} A tooth that fails to erupt in its normal functional position by the time it generally should, while its tooth bud has presented in the alveolar bone during the developmental stage, may be considered impacted.3 Tooth impaction refers to a situation where failure to erupt is apparently due to mechanical blocking and consequently the tooth does not erupt, even after the normal time of eruption, while adjacent teeth have already properly erupted.^{4,5} Cases in which all teeth have begun to erupt but there is a generalized delay, should be termed a delayed eruption and not an impaction.6 An impaction may be primary, namely the tooth has never erupted (also called primary failure of eruption), or it may be secondary, mean-

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Phone no.: 972-4-8542265 Fax no.: 972-4-8339889 E-mail: aizenbud@ortho.co.il ing that the tooth is re-impacted after it has erupted.⁷ The majority of impacted teeth are permanent teeth and are relatively common at the early mixed dentition age.⁸ Furthermore impaction is rare in primary dentition, and in most cases involves the first and second molars.⁸ Impaction of primary anterior teeth is extremely rare and has been occasionally reported in the literature.^{3,7,9-12}

Impacted teeth tend to be asymptomatic and may be discovered by means of routine radiographic examination. Impaction is caused by systemic or local etiological factors. Local factors associated with impacted primary teeth include: Odontomas, odontogenic tumors, ankylosis, traumatic injuries and dentigerous cysts. Impaction can also be the result of lack of the space necessary in the dental arch for normal eruption, supernumerary teeth, malposition and malformation of tooth buds or infectious processes in the eruption path.1 One of the most common reasons for tooth impaction is odontomas. These are developmental anomalies resulting from the growth of completely differentiated epithelial and mesenchymal cells that give rise to functional ameloblasts and odontoblasts. These cells in turn form variable amounts of enamel, dentin and pulpal tissue of the odontoma.13

This report presents a case of a 5.5 year-old boy with an impacted malformed primary central incisor, which never erupted in the oral cavity, and was histologically diagnosed as a compound odontoma. The clinical as well as radiographic diagnoses are discussed and alternative management treatment procedures are presented.

CLINICAL CASE REPORT

A 5.5 year-old Israeli boy was referred to the pediatric

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Figure 1. Intraoral frontal view of the maxilla and the mandible when the child was first admitted to the pediatric dental clinic. The space of the missing maxillary left primary central incisor was naturally maintained with no midline deviation, and the buccal swelling was palpated.

dental clinic at the Rambam Medical Center, Haifa, Israel, due to a delayed eruption of a maxillary primary central incisor. His medical history revealed uneventful pregnancy, normal delivery and no systemic diseases. No history of dental trauma or infection was reported. Intra-oral examination showed a full complement of primary teeth with the exception of the upper left primary central incisor. A mild gingival buccal swelling was palpated in the area of the unerupted incisor, which included a tooth bud (Figure 1). The space for the eruption of the maxillary left primary central incisor was naturally maintained in the dental arch and no midline deviation was diagnosed in comparison to the mandibular dental arch and the facial midlines (Figure 1). Bilateral class I occlusion was observed in the area of the primary canines, while the second primary molars on both sides exhibited mesial-step occlusion. Periapical radiographs taken from two different angles (Figure 2a and 2b) revealed a radio-opacity tooth-like conglomerate mass with a partially developed root and a malformed crown in the area of the unerupted maxillary left primary central incisor. The delayed development of the successor maxillary left permanent central incisor in its eruptive path in comparison to the right central incisor was noted (Figure 2a).

Based on the clinical examination and radiographic records, a diagnosis of an impacted, malformed primary left central incisor was determined, and surgical removal of the impacted primary tooth was recommended. Under intravenous (IV) sedation (propophol 2-4 mg/kg) a labial full-thickness buccal flap was raised in the periapical region between the maxillary right primary central incisor and the left primary lateral incisor (Figure 3). The impacted left upper primary incisor was extracted, the surrounding tooth follicle soft tissue was removed (Figure 4) and a clean socket was achieved. The flap was sutured by means of resorbable Vicryl 4/0 (Ethicon, New Jersey, USA) using simple interrupted sutures. The extracted specimen was then sent for histological evaluation that revealed a tissue structure characteristic of a compound odontoma.

Postoperative clinical and radiological follow up recall examinations revealed complete healing of the area of the left upper incisors after one month. In a periapical radiograph taken 6 months after the surgical procedure (Figure 5), normal development as well as a normal permanent central incisor pattern of eruption was indicated.

DISCUSSION

Impaction is defined in cases where the tooth does not appear in the oral cavity, and may be covered by a thick layer of alveolar bone i.e. bony impaction, or soft tissue of oral mucosa and gingiva in cases of soft tissue impaction.⁴ Primary tooth impaction is rare, but when it occurs it usually involves the first and second molars.^{3,4} In the anterior segments of the alveolar ridges, primary tooth impaction is

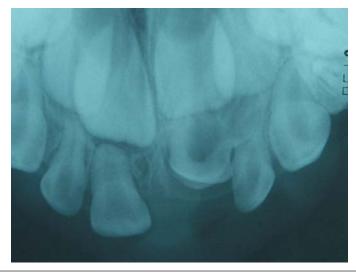




Figure 2a and 2b: Periapical radiography taken from two different angles, reveals an opaque tooth-like mass with a partially developed root in the area of the unerupted maxillary left primary central incisor.



Figure 3. Surgical exposure of the tooth mass.

even rarer. Only a few cases have been reported in the literature.^{3,7,9} Previous reports have described impactions or failure of eruption due to unknown etiology.^{3,7,11}

Ankylosis may create a major etiological factor in cases of primary impaction. In these cases the impacted primary tooth may prevent (often as a physical barrier) the eruption of its successor tooth at its expected time. ¹⁴ Ankylosis might be the result of alterations in the dental follicle or periodontal ligaments covering the tooth bud which leads to a fusion of the cementum to the surrounding bone. The etiology of this process is usually unknown. ^{14,15}

Other factors associated with the interference of normal primary anterior tooth eruption are odontomas. ¹⁶ Odontomas commonly occur in permanent dentition and are rarely reported in association with primary teeth. ¹⁷⁻¹⁹ Katz (1989)²⁰ found only 5 cases (1.26%) of compound and complex odontomas in association with unerupted primary teeth in his analysis of 396 cases of odontomas. In the other reported cases, odontomas were present in the maxillary primary canine area. ^{16,20-23}

Andreasen (1994)²⁴ describes an odontoma-like malformation of the permanent tooth germ due to intrusive luxation or avulsion of the primary tooth. This malformation is rare sequelae to injuries in primary dentition. Reported cases²⁴⁻²⁶ are confined primarily to the maxillary incisors. The histology and radiology of these cases show a conglomerate of hard tissue having an odontoma morphology including separate tooth elements similar to the case we describe in this report. However, Andreasen did not mention this type of malformation as an option of an odontoma-like malformation arising in the primary incisor tooth germ when the permanent tooth bud is normally developed. Furthermore the



Figure 4. Tooth mass and the surrounding tooth follicle soft tissue after removal.



Figure 5. Periapical radiography taken 6 months post operatively, reveals normal development of all permanent incisor tooth buds.

mechanism described by Andreasen is based on a history of a permanent tooth bud pre-eruptive trauma. A vertically directed force through the long axis of the deciduous incisor was transmitted to the permanent tooth germ causing extensive damage. According to this theory the malformation occurs during the early phase of odontogenesis and affects the morphogenetic stages of the ameloblastic development of the permanent tooth germ.

This case report presents a 5.5 year old healthy boy with an unknown history of trauma to the premaxillary region. Clinical and radiographic examinations revealed an asymptomatic impaction of the malformed left primary maxillary central incisor. No evidence of space loss, concomitant asymmetry and disharmony of the maxillary dental arch or occlusal relations were recorded. Careful examination of the periapical radiographs from two different angles (Figs. 2a and 2b) revealed a strange shape including two cusps (premolar-form) of the left primary incisor with only a partially developed root and a large pulp chamber. In this case non eruption of the upper left primary central incisor was diagnosed as a primary failure of tooth eruption. However it was

not clear whether the tooth bud was an odontoma which developed in the site, or a malformation of the tooth bud itself.

The treatment goal in this case was to prevent any damage to the developing permanent tooth, and to allow its normal eruption. Therefore surgical removal of the unerupted primary incisor was considered as the preoperative periapical radiograph revealed a delay in the development of the permanent successor (the left permanent central incisor) in comparison to the right central incisor. Previous reports have described primary failure of eruption in primary teeth that have severely delayed the eruption of permanent dentition.²⁷ However, it has been reported that surgical extractions of impacted teeth have allowed normal eruption of permanent successor teeth without any orthodontic treatment.79 If the roots of the central incisor are completed, the eruption force will be affected and orthodontic correction might be necessary. Some authors prefer to delay surgical removal of the impacted tooth to avoid injury to the developing permanent tooth buds until the permanent root is completed, and then an orthodontic traction can be accomplished.28

In the presented case the surgical intervention was carried out under IV sedation due to the young age of the child, the position of the impacted tooth in the premaxilla and poor cooperation for this surgical procedure. In periapical radiographs taken 6 months post-operatively, normal permanent central incisor tooth bud development and a normal pattern of eruption was observad. The catch up development in the eruption path of the left permanent central incisor when compared with that of the right one, from pre and postoperative periapicals (Fig 2a and 5) may be the consequence of early surgical removal of the primary tooth-like mass.

This mass was preliminarily diagnosed as the tooth bud of a primary central incisor that did not develop properly during the morpho-differentiation stage. The differential diagnosis revealed an atypical odontoma, since it was not developed adjacent to an unerupted primary tooth bud. A dilaceration of the primary tooth bud¹⁷ was ruled out since the parents insisted that they did not recall any traumatic event. However, trauma that occurs before the eruption of the primary teeth in the oral cavity, may harm the development and eruption of the primary dental bud especially of the incisor area. 18,29 Consequently an odontoma-like malformation of the primary incisor tooth germ might also be a potential diagnosis. Nonetheless, Andreasen²⁴ did not mention this type of developmental malformation, which is very similar to our reported dental anomaly of the primary tooth germ, but rather he referred to the permanent one. Although in our case, even if the parents did not report early facial or dental trauma we can speculate that very early injury of the gum pads during the developmental stage of the primary maxillary incisor crown during at a perinatal or neonatal age resulted in odontoma-like malformation of the primary tooth buds with the same possible mechanism described for the permanent one. If so, the vertically directed force through gum pads may have been transmitted to the primary tooth germ causing damage and may also have caused cessation in

the development of the successor permanent tooth germ. However, in this case the deformity was confined to the primary tooth germ whereas the permanent tooth buds developed normally in their eruption path. According to the cases found in the literature, ankylosis could also be a differential diagnosis for this case. Perhaps there was a primary impaction due to ankylosis of the developing tooth crown. This bony-like fusion in the area of the crown may have caused a developmental malformation in the crown and root area during the morphodifferentiation stage, resulting in a tooth–like mass that looks like a tooth. 10,14

The final diagnosis was only given after histological work up, revealing a compound odontoma in the area of the maxillary left primary central incisor. This finding indicates that the tooth bud of the primary central incisor was missing and a tooth like mass i.e. an odontoma developed. The coincidence of an odontoma and a missing tooth is extremely rare. Odontomas are usually found in the area of unerupted permanent tooth buds and rarely in the area of primary central incisors.¹⁸ It is tempting to speculate a causal relationship, i.e. that an unknown local pathology factor may have physically altered the development of the tooth bud of the primary central incisor which led to the development of an odontoma and the delayed eruption. The occurrence of an odontoma together with the missing primary tooth bud or odontoma -like malformation of the primary incisor has not been previously reported in literature, since odontomas are usually found in the surrounding area of an unerupted tooth.

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