

CASE REPORT

Erupted composite odontoma associated with malformed teeth – unusual dental aberrations following maxillofacial trauma

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Abstract

Background: Tooth development may be hampered following traumatic injuries to maxilla or mandible. The developmental deviations that occur in a tooth bud in the line of a jaw fracture range from agenesis to hamartomatous proliferation. Odontomas are benign odontogenic hard tissue tumors, which are hamartomas characterized by slow growth and non-aggressive behavior. 'Erupted odontoma' is a term used to specifically denote odontomas, which are exposed into the oral cavity. These are rare entities with only 25–30 cases being reported so far in the dental literature. **Case Description:** This clinical report is pertaining to a rare case of an erupted odontoma in an adolescent patient which was presenting as a hard tissue outgrowth along with an extra-oral sinus in the posterior mandibular region. The surgical excision of the odontoma along with the associated malformed teeth resulted in resolution of pain and sinus discharge associated with this lesion. **Conclusions:** Erupted odontomas need to be considered in the differential diagnosis of hard tissue projections or outgrowths in the site of past maxillofacial injuries. Surgical excision of these lesions enable resolution of patient symptoms associated with this condition as evidenced in the present case and in other cases cited in the literature. The erupted odontoma attached to malformed molars and presenting with signs and symptoms suggestive of osteomyelitis is a rare combination of developmental aberrations following maxillofacial injury.

Keywords: odontoma, trauma, maxillofacial, mandible, osteomyelitis, malformation.

Introduction

Traumatic injuries to tooth bearing portions of the jaw may abnormally alter or arrest the development of teeth in the line of injury. Numerous longitudinal studies have been performed to assess the fate of developing tooth buds in the line of jaw fractures. The common abnormalities in these teeth include – hypoplastic defects, crown and/or root dilacerations, deficient root formation, structural abnormalities of root, obliteration of the pulp cavity, impaction, odontoma-like malformations, external resorption and eruption disturbances [1, 2].

Odontomas are hamartomas of aberrant tooth formation, which occur due to budding of extra-odontogenic epithelial cells or detachment of odontogenic cells from dental lamina [3]. These odontogenic cells may in turn differentiate and deposit enamel, dentine, cementum or pulp in the form of multiple teeth like structures (compound odontoma), amorphous calcified masses (complex odontoma) or a combination of both (composite odontoma and compound-complex odontoma). Trauma to the tooth bud during its early developmental stages has been proposed as a possible predisposing factor for the origin of odontoma [4, 5].

Odontomas erupting into the oral cavity has been considered a rare phenomenon. The first case of an erupted odontoma was reported by Rumel A *et al.* in 1980 [6]. Ever since then, approximately 25 cases of erupted odontomas have been reported. This exposure of the odontoma into the oral cavity has been reported to be associated with numerous symptoms like pain, swelling, facial asymmetry and recurrent infections [7].

We report a unique case of an erupted composite odontoma, possibly occurring as sequelae to trauma and resulting in chronic osteomyelitis.

Patient, Methods and Results

A 12-year-old boy of Indian origin reported to the Department of Pediatric Dentistry, C.S.M. Medical University, Lucknow, India, with a complaint of painful swelling on the left cheek region of 10 days duration (Figure 1a). The patient was of normal height and weight and his medical history was non-contributory. However, the patient reported that he met with a trauma before four years, which resulted in 'severe bleeding in the same region and difficulty in mouth opening' for which he

underwent treatment at a local hospital. The patient's past treatment records were not available for perusal, but from the parent's narration, it was discerned that he underwent intermaxillary fixation for three weeks.

Clinical examination revealed the presence of a well-defined oval soft swelling measuring about 4 cm in diameter

in the left cheek region, 3 cm above the lower border of the mandible. An extraoral sinus with active pus discharge was observed in the centre of the swelling. Intraorally, the left mandibular second molar was missing and in its place, a yellowish-brown hard, tender mass measuring 6×13 mm, was observed (Figure 1b).

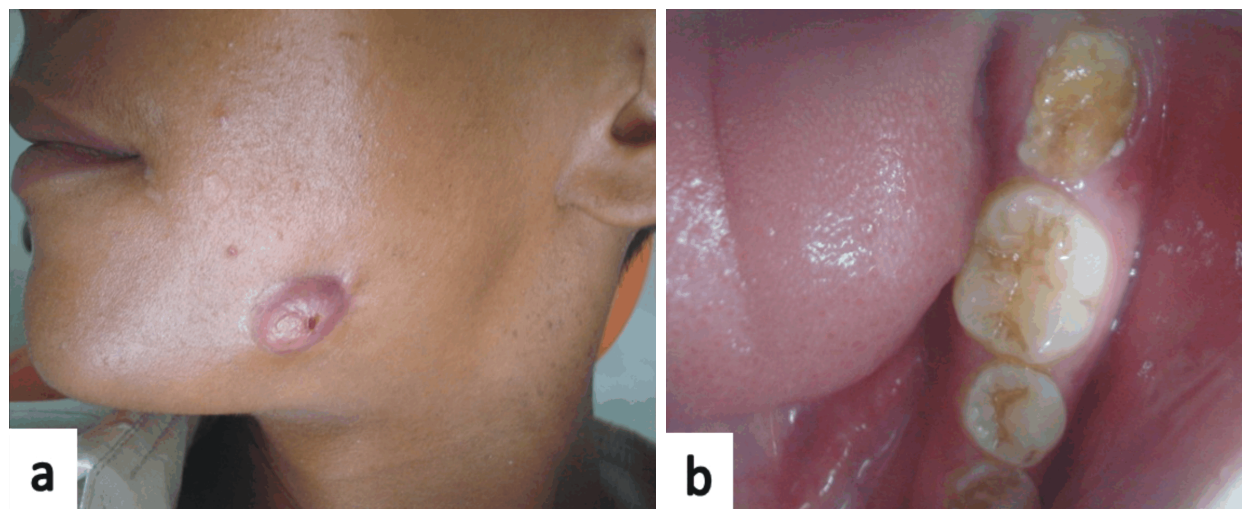


Figure 1 – Extraoral sinus with active pus discharge in the left cheek region (a) and hard tender mass in the corresponding region intraorally (b) present instead of mandibular second molar.

On radiographic examination, this hard tissue lesion was found to be well defined, radiopaque, uniformly dense and separated from the surrounding bone, suggestive of a sequestrum. The mandibular second and third molars were impacted beneath this radiopaque mass. The crown portions of these impacted teeth appeared to be united but, the root portions were distinct and dilacerated. A radiolucent zone was observed apical to and along the distal border of these incompletely fused teeth, which was suggestive of inflammatory bone resorption (Figure 2).

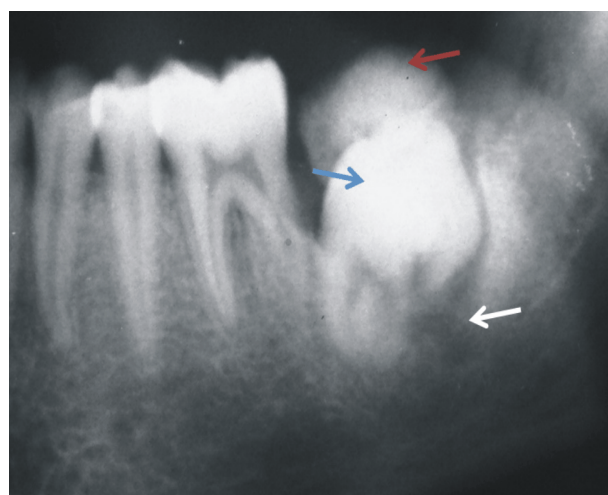


Figure 2 – Radiovisiographic view showing intraoral hard tissue lesion (red arrow), fused molars (blue arrow) and radiolucency apical to the fused molars (white arrow) suggestive of inflammatory bone resorption.

Considering the similarity between the density of the radiopaque mass and the surrounding bone (sequestrum), presence of irregular radiolucent zones of bone destruction coupled with an extra-oral sinus in the adjoining region;

a provisional diagnosis of chronic osteomyelitis secondary to folliculitis in relation to impacted 2nd and 3rd molars was made.

An empirical antibiotic regimen consisting of Amoxicillin along with Clavulanic Acid and Metronidazole was initiated along with analgesics (Diclofenac Sodium).

The hard tissue mass and the malformed teeth were surgically excised as two separate masses through an intraoral approach under local anesthesia.

The underlying necrotic bony margins were curetted and the surgical site was filled with alloplastic bone graft (Bio-Oss[®], Geistlich Pharma AG, Wolhusen, Switzerland). Microbial culture of the curetted segments predominantly revealed the presence of streptococcal organisms, which were sensitive to penicillin group of drugs.

Gross examination of the excised specimen revealed that the excised left mandibular 2nd and 3rd molars were invested within a fibrous band. The mandibular 2nd molar was relatively short with well defined crown and root portions, while the mandibular 3rd molar had poorly developed crown and roots (Figure 3a).

A thin ground section of the overlying hard tissue mass was prepared and subsequent histopathological examination of this section revealed disorganized conglomerates of enamel and dentine, which was suggestive of a complex odontoma (Figure 3b).

Considering the clinical, radiological and histopathological results, a diagnosis of chronic osteomyelitis associated with an erupted composite odontoma was made.

The patient was reviewed after one week, six months and one year. After one year, the extra-oral sinus was seen to heal well without any signs of scarring (Figure 4a).

Postoperative radiographs taken after one year revealed excellent healing of the surgical site (Figure 4b).

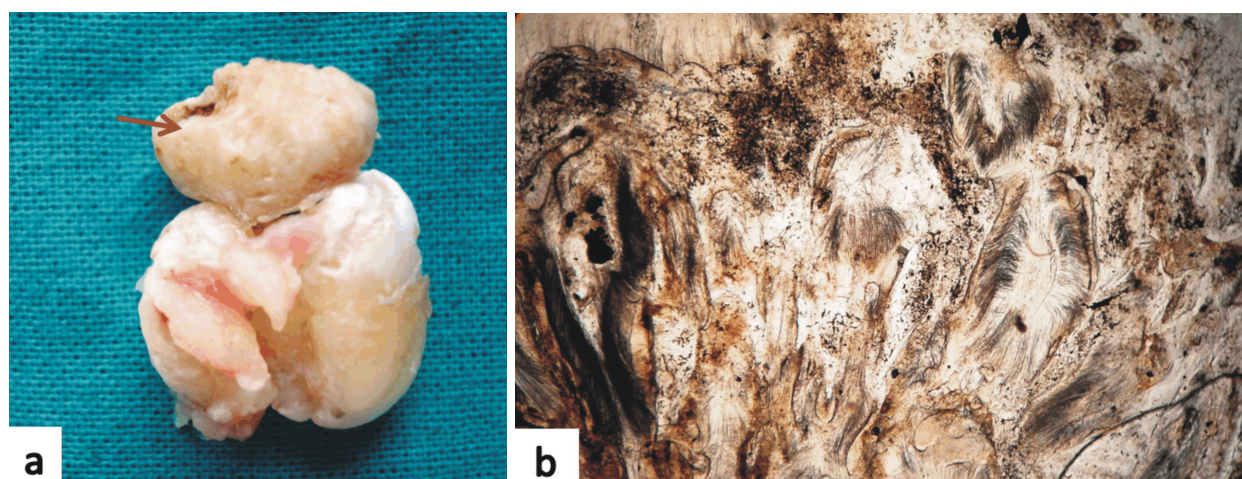


Figure 3 – Surgically excised left mandibular 2nd and 3rd molars (a) which were linked by a fibrous band along with the erupted odontoma (red arrow). Ground section of the erupted odontoma (b) revealing disorganized conglomerates of enamel and dentine.

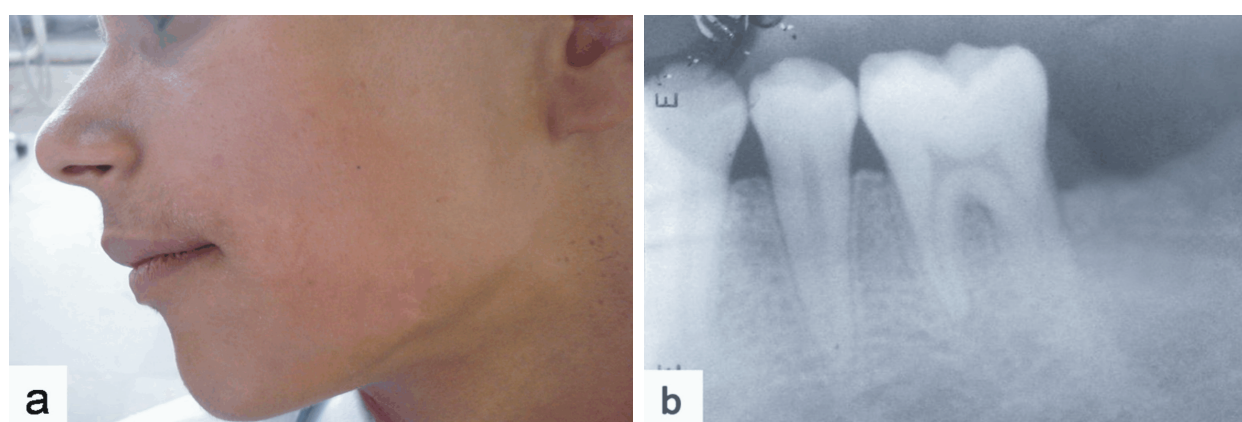


Figure 4 – Follow-up after six months (a) revealing minor scar at the site of extraoral sinus and radiographic view after one year (b) depicting healing of the surgical site.

Discussion

Traumatic jaw injuries along the sites of active odontogenesis can negatively influence the morphological and histological structure of the tooth in the line of injury. Infection, rotation/displacement of tooth bud, and contact between the developing tooth bud and agents used for fixation such as intra-osseous wires, screws, etc. have been reported to be associated with arrested growth and other abnormalities [1, 2]. The relationship between the occurrence of dental abnormalities and the type of fracture is inconclusive.

Odontoma-like malformations of successional (permanent) teeth have been commonly reported following intrusive and avulsive injuries to primary teeth [8–10]. However, odontoma-like malformations of accessional teeth following injuries/fractures of tooth bearing portions of jaws are relatively rare. Development of compound and complex odontomas in experimental animals following traumatic injuries of jaws have been documented by Glasstone S [4] and Levy BA [5] respectively. The results of a literature search on odontomas associated with jaw fractures/injuries revealed very few case reports.

Güngörmüş M *et al.* reported a case of compound odontoma, which had formed in place of right mandibular 3rd molar in a patient who had been treated for a mandibular fracture in the left (contralateral) parasymphysis

region by means of open reduction and plate fixation [11]. An associated abnormality was observed in the distal root of the right mandibular 2nd molar which had deficient root formation. No fracture lines were evident in the mandibular posterior region and the authors inferred that the injury to the parasymphyseal region would have indirectly affected the mandibular posterior region of the opposite side. However, in the present case, the patient provided history of a direct blow to the mandibular posterior region.

Ferguson MW and Whitlock RI reported a case of complex composite odontoma in the mandibular 3rd molar region in a patient with unilateral condylar hypoplasia, who met with a condylar fracture at an early age [12]. Recently, Hammoudeh JA *et al.* reported a case of bilateral complex odontoma in the posterior mandibular region in a 4-year-old girl, which was attributed to the placement of external mandibular distractors during the neonatal period [13]. In all these reports, odontomas were observed in the posterior mandibular region following direct or indirect injury to this region.

In the present case, the origin of composite odontoma can be attributed to the traumatic episode, which could have resulted in displacement of the developing right mandibular 3rd molar tooth bud and budding or detachment of differentiated odontogenic tissues from mandibular 3rd and 2nd molar enamel organ. These well-differentiated extra-odontogenic tissues would have given rise to functional

ameloblasts, odontoblasts and cementoblasts, which would have in turn resulted in deposition of enamel, dentine and cementum in an abnormal manner. Displacement of the mandibular 3rd molar tooth bud could have occurred even before the morphodifferentiation stage of mandibular 3rd molar (when the mandibular 2nd molar's crown formation would have been completed) resulting in relatively greater disfigurement of the crown and root portions of the 3rd molar (when compared to the 2nd molar).

Increase in the size of odontoma, remodeling of the surrounding bone, reactive growth of the capsule and forces transmitted by the underlying impacted tooth have been hypothesized as factors resulting in 'eruption' of odontoma [14]. Secondary infections around an erupted odontoma have been attributed to the lack of adhesion between the odontoma and the surrounding soft tissue/bone interface resulting in increased chances of microbial invasion. Bertolai R *et al.* reported a case of Ludwig's angina associated with an erupted odontoma in a 29-year-old female, which was managed by antibiotic therapy, intraoral incisions to drain the swelling and surgical excision of the odontoma [15]. In the present case, the erupted odontoma was associated with pain, extraoral sinus formation and radiolucencies in the surrounding bone, which was suggestive of chronic suppurative osteomyelitis. Antibiotic therapy associated with surgical excision of odontoma, surgical curettage of underlying bony margins and alloplastic bone grafting enabled resolution of the extraoral sinus and good healing of the bony region distal to the mandibular first molar.

The components of an odontoma are usually held within a capsule of peripheral fibrous connective tissue, which actually represents the dental follicle or periodontal ligament of abnormal dental structure [3]. Encapsulation of the dental components within this fibrous capsule enables easy enucleation of the odontoma as it clearly demarcates odontoma from the surrounding bone. In the present case, the malformed molars were united by means of a fibrous band, which enabled surgical excision of the malformed molars with relatively greater ease.

Odontomas are the most common types of odontogenic tumors, the prevalence of which exceeds the combined prevalence of all other odontogenic tumors put together [3]. But, exposure of an odontoma ('eruption') into the oral cavity has been considered as a rare phenomenon by many authors [6, 7, 15]. This fact can be considered to be partially true if we eliminate supernumerary teeth (which are essentially compound odontomas) from this category. In a recent review pertaining to erupted odontomas, the authors found that out of the 20 reported cases of erupted odontoma in dental literature, nine were compound odontomas and 11 were complex odontomas [7]. The present case can be considered unique, due to scarcity of reports pertaining to erupted composite (compound-complex) odontomas and due to its possible association with maxillofacial trauma in a young patient.

Conclusions

Erupted odontomas are rare entities, which need to be considered in the differential diagnosis of malformed hard tissue projections in oral mucosa. The present case highlights the diagnosis and management of this condition and also emphasizes the probable role of maxillofacial trauma in the origin of this condition. The combined occurrence of malformed and dilacerated molars, and erupted odontoma; is an interesting combination of embryological aberrations occurring in a site of injury, and the identification of the same following the occurrence of chronic osteomyelitis is a rare entity.

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