Management of compound odontoma in a 10-year-old girl preserving the associated impacted permanent tooth

ABSTRACT

Background Compound odontoma has been reported to be the most common of all odontogenic neoplasms and tumor-like lesions. Only rarely the treatment of this lesion in association with an impacted tooth has been reported.

Case report A compound odontoma in a 10-year-old girl, associated with an impacted permanent incisor is described, focusing on the diagnosis and the importance of early treatment of this lesion. The patient underwent surgical excision of the lesion and it was decided to wait for the spontaneous eruption of the impacted tooth. After 6 months no eruption was observed and thus the orthodontic treatment was deemed necessary. At the one-year follow-up, the tooth was brought into the maxillary arch.

Keywords Compound odontoma; Impacted tooth; Orthodontic therapy.

Introduction

Odontoma, compound type (OCp), is a tumor-like malformation (hamartoma) with varying numbers of tooth-like elements (odontoids) [Praetorius and Piattelli, 2005]. Clinically, OCps are painless and slowly growing lesions. However, an affected patient may present pain when a permanent tooth or multiple teeth fail to erupt [Nagaraj, Upadhyay, Yadav, 2009]. The lesion is composed of tissues native to teeth, such as enamel, dentin, cementum and pulp. Odontomas as further sub-classified upon their gross and radiographic features into compound (small tooth-like structures) or complex (a conglomerate of dentin, enamel and cementum) [Bordini Jr et al., 2008]. According to the WHO definition, odontoma is a congenital developmental defect, resulting from growth of completely differentiated epithelial and mesenchymal cells, in which all kinds of dental tissues can be found. In a study it was underlined that the bulk of the tumour was formed of dentin and highly of histo- and morphodifferentiated tissues [Piattelli, Trisi, 1992].

Similar to teeth, once fully calcified, they do not develop further [King, Wu, 2002]. Odontomas are usually diagnosed in the first and second decade of life [Praetorius and Piattelli, 2005; Owens et al., 1997]. In a study by Amado Cuesta et al. [2003] the mean age of the patients was 18 years with the majority of odontomas occurring in the first (32%) and second decade (38%) of life. Tomizawa et al. [2005] described that 50% of odontomas were found in the first decade of life and that the youngest patient was 14 months old. Katz [1989] reported that odontomas rarely involved the primary dentition and found only 5 out of 396 odontomas (2%) associated with primary teeth, whereas these lesions were most commonly removed from the 11-15-year-old age group, and they were apparently age- and site-related. Specifically, those in the incisal area were diagnosed and treated at an earlier age than those in the canine or third molar regions [Katz, 1989; Delbem et al., 2005].

Regarding the location, odontomas are most commonly found in the maxilla (70%), and particularly, the anterior maxilla seems most frequently affected (62%). Interestingly, both types of odontoma occurred more frequently on the right side of the jaw than on the left (compound 62%, complex 68%) [Amado Cuesta et al., 2003; Kaugars et al., 1989; Regezi et al., 1978; Cawson, Odell, 1998].

There are conflicting evidences in the literature regarding the gender predilection: some studies have found that males are more likely than females to have odontomas, while other studies reported that females are more frequently affected (52.4%) [Amado Cuesta et al., 2003]. According to Praetorius and Piattelli [2005], OCps may occur in any tooth-bearing area of the jaw with no gender predilection.

The precise aetiology of odontomas is unknown, but local trauma, infection, inheritance, and genetic mutation have been postulated as possible causes [Owens et al., 1997].
Odontomas can be associated with an impacted tooth and occasionally with a dentigerous cyst [Motokawa et al., 1990; Gallana-Alvarez et al., 2005; Gigliotti et al., 1975]. Chang et al. [2003] reported that 64 (79%) of 81 odontomas were associated with 80 impacted teeth, including 71 permanent teeth, 2 deciduous teeth, and 7 supernumerary teeth. Of the 71 impacted permanent teeth, the maxillary central incisor (27%) was the tooth most commonly affected, followed by the maxillary canine (26%) and mandibular canine (24%).

OCps can also cause displacement of erupted teeth or can occur in a site where the permanent tooth is missing [Praetorius and Piattelli, 2005]. Loose, myxoid connective tissue with odontogenic epithelial rests may be seen in close association with the lesion, and most often this connective tissue represents normal dental follicular tissue. Fibrous connective tissue with a cystic lining representing a dentigerous cyst may also be seen [Nelson and Thompson, 2010].

The choice of treatment is conservative surgery with local excision and the prognosis is excellent. No recurrence of odontomas was found after surgical excision with a follow-up of 1 to 15 years. However, preserving the associated impacted teeth may make complete removal of the lesion difficult and may explain some cases of recurrence [Silvia et al., 2007].

We report a case of odontoma in a 10-year-old girl, associated with an impacted permanent tooth, focusing on the diagnosis and the importance of early treatment of these lesions.

**Case Report**

A 10-year-old Caucasian girl, presented to the Pediatric Dentistry division of the Dental School, University of “G. D’Annunzio” of Chieti-Pescara for the eruption disturbance of a permanent central incisor tooth (tooth 2.1).

The medical history of the patient was non contributory, and the clinical examination revealed a localised, hard, swelling lesion, located in the upper left anterior region of the maxilla, covered by normal mucosa and gingiva (Fig. 1). No pain was referred. The upper right central and lateral incisors had already erupted without complication. Periapical and panoramic radiographs revealed multiple radiopaque tooth-like structures surrounded by radiolucent areas around the crown of the unerupted tooth (Fig. 2).

Surgical excision of the odontoma was performed. Macroscopically, the specimen was composed of 7 small calcified structures encapsulated with fibrous connective tissue (Fig. 3). A clinical diagnosis of compound odontoma was made. The lesion was then sent for histological examination, which confirmed the clinical diagnosis.

Microscopically, the odontoma showed immature enamel, dentin and pulp with no root formation. In the connective tissue capsule surrounding the odontoma, a significant number of odontogenic epithelial islands were observed (Fig. 4) [Mummolo et al., 2010].

Regarding the treatment of the impacted tooth, as the root apex closure was still incomplete it was
decided to wait for spontaneous eruption. However, 6 months after surgery an orthodontic treatment was deemed necessary, and thus a palatal arch and a direct-bonding system were applied (Fig. 5). Follow-ups were scheduled at monthly intervals.

At the one-year follow up, the tooth was brought into the maxillary arch (Fig. 6).

Discussion

A tooth is impacted when eruption into a normal functional position is obstructed by a physical barrier [Yeung et al., 2003, Motokawa et al., 1990]. Causes of dental impaction include developmental anomalies, such as malposition, dilaceration, ankylosis; systemic or genetic diseases, such as cleidocranial dysostosis and hypopituitarism; tumors, odontoma, dentigerous cysts, and presence of supernumerary teeth [Snawder, 1974; Motokawa et al., 1990, Yeung et al., 2003].

In the present report the cause of dental impaction was a compound odontoma. Indeed, odontoma is frequently discovered due to impaction of the permanent teeth with or without persistence of the primary teeth or, less frequently, it is an accidental radiographic finding [Owens et al., 1997; Katz, 1989].

Impacted teeth do not always erupt spontaneously after removal of odontomas. In a study by Ashkenazi et al. [2007] the effect of several variables on eruption of impacted teeth in children with supernumeraries or odontomas was examined. Loss of space, a second surgical procedure, a third surgical procedure, and orthodontic treatment were recorded in 77.6%, 53.8%, 9.4%, and 85% of the patients, respectively. Spontaneous eruption occurred in 83%, 75%, 46%, 19%, and 32% of the impacted teeth with normal and small size, conical, tuberculated, and odontoma forms, respectively. In the univariate analysis, spontaneous eruption correlated with apex distance of the impacted tooth relative to its estimated position (P <0.001), extent of vertical impaction (P <0.001), obstacle form (P <0.019), stage of root development of supernumerary tooth (P = 0.006), angle of impaction relative to the midline (P =0.015), and time of surgery (P =0.05). In the multivariate logistic regression analysis, higher distraction of the apex of the impacted tooth relative to its estimated correct position and the obstacle form (tuberculated and odontomas) were independently associated with impediment of spontaneous eruption (P =0.03 and P =0.04, respectively). The Authors concluded that spontaneous eruption of impacted teeth correlated mostly with lower distraction of the impacted tooth apex and obstacle form (conical and superlative) [Ashkenazi et al., 2007].

Immediate orthodontic traction is recommended concomitantly with the first surgery to remove supernumerary teeth [Ashkenazi et al., 2007].

However, opinions differ concerning the optimal time for surgical intervention. Some studies recommend removal soon after diagnosis to prevent loss of eruption potential, loss of space and midline shift, and more extensive surgical and orthodontic treatment for correction [Kokich and Mathews, 1993; Motokawa et al., 1990; Snawder, 1974; Brunetto et al., 1991; de Oliveira et al., 2001].

In our case there was still a residual eruption potential
due to the incomplete closure of the incisor root apex. For this reason it was decided to wait six months to radiographically evaluate any spontaneous eruption movements. However, since the tooth did not appear to move, a second surgery was performed and an orthodontic traction was applied as done in another work by Yadav M. et al. [2012].

Indeed, 55% of the impacted teeth (sum of published data, 104 out of 188) need orthodontic treatment because of space loss or ectopic eruption. Thus, in the present case it was worth waiting and trying to avoid the orthodontic treatment. Moreover, orthodontic therapy is not usually applied to guide the impacted tooth into a normal position after excision of compound odontomas because most odontomas are smaller than a normal tooth and the impacted permanent tooth is not usually highly deviated [Kamakura et al., 2002; Veis et al., 2000; Torreti and Carrel, 1983]. In general, if the root of the permanent tooth is still developing, the tooth may erupt normally; however, once the root apex has closed, the potential to erupt is drastically reduced or lost, and thus the orthodontic therapy is necessary [Kokich and Mathews, 1993; Kamakura et al., 2002].

Acknowledgements

The Authors would like to thank Dr. Marcello Piccirilli (Dental School, University of Chieti-Pescara, Italy) for the technical support in the histology processing. This work was partially supported by the National Research Council (C.N.R.), Rome, Italy, by the Ministry of Education, University, Research (M.I.U.R.), Rome, Italy.

References