Timing and treatment sequence in the management of odontomas associated with impacted teeth: A literature review and report of two cases

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Abstract

Aim This study aims to highlight the importance of early diagnosis, timing, optimal treatment sequence and multidisciplinary approach as key factors in the orthodontic management of impacted and retained teeth associated with odontomas.

Methods Literature about classification, epidemiology, aetiopathogenesis, histopathology and therapeutic options about odontomas and impacted teeth in orthodontics was reviewed. Two case reports are presented, showing different timing in diagnosis and surgical removal of odontomas and some biomechanical approaches.

Results Optimal treatment sequence, multidisciplinary approach, early diagnosis and timing are key factors in orthodontic and surgical-orthodontic management of impacted and retained teeth associated with odontomas.

Conclusion An early removal of the odontoma is certainly a more effective and simpler procedure in the approach to this problem.

KEYWORDS Odontoma, Impacted teeth, Tooth retention, BMP4, Wnt signaling pathway, R-Spondin3, Wnt/β-catenin, Podoplanin

Introduction

This study aims to highlight the importance of early diagnosis, timing, optimal treatment sequence and multidisciplinary approach as key factors in the orthodontic management of impacted and retained teeth associated with odontomas. Definition, classification, epidemiology, aetiopathogenesis, literature review and treatment sequences will be discussed by means of two clinical case reports.

Definition and classification

According to 2017 WHO Classification of odontogenic tumors and cysts [Soluk-Tekkeşin and Wright, 2018] (modified, Table 1), odontoma is included as a benign odontogenic tumor of mixed origin (epithelial-mesenchymal) [Gyulai-Gaál et al., 2007], characterised by the presence of odontogenic tissues (enamel, dentin, cement) and cells (ameloblasts and odontoblasts) in a well differentiated form and composing calcified masses. The lesions are defined as compound odontoma if resembling teeth or a collection of small tooth-like structures (denticles), while in complex odontoma all tissues occur distributed irregularly in a disordered pattern [González-Alva et al., 2011; Kapadia et al., 2007; Mostowska et al., 2003; Soluk-Tekkeşin and Wright, 2018; Wood and Goaz, n.d.]. Odontomas are historically and still considered as a developmental anomay or tumor-like malformation (hamartoma of tooth forming tissue or tooth germ or "dysontogenetic dysplasia").

Description and epidemiology

Odontoma is the most common odontogenic tumor of the oral cavity, accounting for 60–70% of all odontogenic tumors [Paoloni et al., 2012; Wood and Goaz, n.a.], in spite of different data presented by various authors. Odontomas are relatively more frequent in the Caucasian population than the in Asian and African ones [Paoloni et al., 2012], but different standards of dental care and diagnostic protocols probably play a role. In most studies and meta-analyses different data in distribution between male and female population and between maxilla and mandible location are shown, while the presence in anterior regions seems more probable. Da Silva et al. [2019] reported also odontomas as more frequently detected in white males in their second decade, prevailing Complex Odontomas in the maxilla (83,3%) and Compound Odontomas in the mandible (60%). Lower canines were the most frequently odontoma-associated impacted teeth, followed by upper central incisor and upper canine [Aizenbud and Front, 2008; Baldawa et al., 2011; Bayram et al., 2007; Fomenko et al., 2020; Khan et al., 2014; Miki et al., 1999; Nagaraj et al., 2009; Nogueira, 1977; Shelton et al., 1997; Shetty et al., 2013]. Differently, Bereket et al. [2015] reported a female predilection with compound odontoma more frequent than complex odontoma. Similar non-uniformity of data are reported in other studies [An et al.,

2012; Iatrou et al., 2010; Otsuka et al., 2001; Tomizawa et al., 2005; Tuczyńska et al., 2015].

Aetiopathogenesis and histopathology

Unknown or not well-determined causes and many other factors in the process of being identified in tooth eruption and craniofacial morphogenesis are involved in the aetiopathogenesis of odontomas. In a study [Papagerakis et al., 1999] about phenotypic markers of ameloblasts and odontoblast cells, was showed in odontogenic tumors (odontomas, ameloblastic fibroma and ameloblastic fibro-odontomas) an over-expression of genes coding for proteins involved in biomineralisation (amelogenins, keratins, collagen type III and IV, vimentin, fibronectin, osteonectin and osteocalcin) and particularly an interesting co-expression of amelogenins and osteocalcin in the epithelial zone, showed by immunostaining (in situ hybridisation, transitional cells). An important role is attributed to neurotrophic factors, growth factors, transcription factors, various types of signal receptors, signaling molecules and local factors or morphogens regulating skeletal morphogenesis and odontogenesis [Kapadia et al., 2007]: Bmp4 signaling pathway (tooth specific) [Kapadia et al., 2007; Russel et al., 1998], Wnt signaling pathway and modulator R-Spondin3 [Alhazmi et al., 2020], Wnt/β-catenin [Fujii et al., 2019], Podoplanin [González-Alva et al., 2011], epithelial-mesenchymal cell to cell inductive interactions [Papagerakis et al., 1999], anomalies of gubernaculum dentis, inheritance, genetic mutations and inflammatory or infective or traumatic factors that are involved in aetiopathogenetic processes [Fujii et al., 2019; Hainline-Raez and Richardson, 1985; Papagerakis et al., 1999; Russel et al., 1998; Soluk-Tekkeşin and Wright, 2018]. Based on these considerations and on clinical, epidemiological and immuno-histochemical data, an interesting hypothesis has been formulated about the common origin of odontomas and supernumerary teeth, in spite of a distinct classification [Pippi, 2014] . Similarly, genes affecting tooth morphogenesis (homeo-domain protein MSX1 and paired-domain transcription factor PAX9) may be implicated in syndromic and non-syndromic tooth anomalies and agenesis [Kapadia et al., 2007; Mostowska et al., 2003].

Malignant odontogenic tumors	odontogenic carcinomas	ameloblastic carcinoma primary intraosseous carcinoma, not otherwise specified sclerosing odontogenic carcinoma clear cell odontogenic carcinoma ghost cell odontogenic carcinoma
	odontogenic carcinosarcoma	
	odontogenic sarcomas	
Benign odontogenic tumors	Benign epithelial odontogenic tumors	Ameloblastoma: - ameloblastoma, unicystic type - ameloblastoma, extraosseous/peripheral type - metastasizing ameloblastoma Squamous odontogenic tumor Calcifying epithelial odontogenic tumor Adenomatoid odontogenic tumor
	Benign mixed epithelial and mesenchymal odontogenic tumors	Ameloblastic fibroma Primordial odontogenic tumor Odontoma: - Compound - Complex - Compound-complex Dentinogenic ghost cell tumor
	Benign mesenchymal odontogenic tumors	odontogenic fibroma odontogenic myxoma/ myxofibroma cementoblastoma cemento-ossifying fibroma
Odontogenic cyst	odontogenic cysts of inflammatory origin	radicular cyst inflammatory collateral cysts
	odontogenic and non-odontogenic developmental cysts	dentigerous cyst odontogenic keratocyst lateral periodontal cyst and botryoid odontogenic cyst gingival cysts glandular odontogenic cyst calcifying odontogenic cyst orthokeratinized odontogenic cyst nasopalatine duct cyst

TABLE 1 2017 WHO classification of Odontogenic tumors [Soluk-Tekkeşin and Wright, 2018], modified.

Odontomas and clinical orthodontics

A multidisciplinary treatment approach is often required, since odontomas may cause impaction, anomalous eruption, tooth retention or other various anomalies in tooth eruption (delayed, deflection, transposition) or malformation and resorption of neighboring teeth [Altay et al., 2016; Kämmerer et al., 2016; Morning, 1980]. Odontomas are mainly asymptomatic, showing a slow growth and benign behaviour; sometimes they can be appreciated as a palpable dimensional increase at the level of vestibular bone corticals, although they are often located in close proximity to these bony structures [Atarbashi-Moghadam et al., 2019; Cozza et al., 2003; Machado et al., 2015]. This is the reason why most odontomas are detected incidentally at routine radiographic examinations [Kämmerer et al., 2016; Maltagliati et al., 2020], and often in mixed and secondary dentition and in the second decade of life [Batra et al., 2004; Cozza et al., 2004; de Oliveira et al., 2001; Kamakura et al., 2002; Lacarbonara et al., 2017; Lazzati et al., 1991; Tamásy, 1972; Tripodi et al., 2012]. In many cases parents or patients detect the delayed eruption of a permanent tooth or permanence of a primary tooth, in such cases surgical and orthodontic treatment of the impacted tooth is often difficult or challenging. The detection of odontomas in primary dentition or early mixed dentition is quite rare, but when it occurs the timely surgical removal may lead to spontaneous and normal eruption of the permanent teeth without any orthodontic intervention or extractions, while in many cases surgical-orthodontic therapy is a common approach [Altay et al., 2016; Ashkenazi et al., 2007; Bansal et

al., 2014; Brunetto et al., 1991; Conti et al., 2012; Dukić et al., 2007; Jung et al., 2016; Kämmerer et al., 2016; Kupietzky et al., 2003; Liu et al., 1997; Motamedi et al., 2008; Motokawa et al., 1990; Pavoni et al., 2013; Troeltzsch et al., 2012]. Similar results were obtained by Morning et al. [1980] in a retrospective study, with about 75% of spontaneous eruption of impacted teeth related to odontomas.

In a particular stage of development and before planning orthodontic alignment after removal of an odontoma, it may be indicated to wait for spontaneous eruption (six months or until complete formation of the root of the impacted tooth or homologous tooth erupted with complete root formation for at least six months) [da Costa et al., 2008]. Despite sometimes the natural potential of tooth eruption is retained, ortohodontic treatment is still required for mantaining or creating space in the dental arch [Berthold et al., 1987; Lorber, 1977]. Monitoring of the periodontal health and stability of disimpacted teeth in the long term is also required [Nagaraj et al., 2009].

After initial detection of odontomas by panoramic radiographs, Cone Beam Computed Tomography (CBCT) [Chaushu et al., 2004; Gurler et al., 2017; Kobayashi et al., 2013; Troeltzsch et al., 2012] is the method of choice for the diagnosis and planning of surgical and orthodontic treatment of impacted teeth (both by supernumerary teeth and odontomas), especially to avoid damage to the neighbouring teeth and anatomic structures [Jung et al., 2016].

Autotrasplantation is another more recent method based on the traditional surgical-orthodontic treatment of teeth impacted due to odontomas, which satisfies an immediate cosmetic demand, and may also lead to proper correction of bone defects [Hwang et al., 2017; Robindro Singh et al., 2015].

Case report 1

A healthy 9 years and 6 months old male patient underwent a routine dental examination, not showing intraorally and extraorally any noteworthy problems. Panoramic radiographs (Fig. 1) suggested the presence of an odontoma in the anterior left mandibular region, hindering eruption of tooth 3.2 and probably also of tooth 3.3. Therefore, it was decided the early removal of the odontoma, to allow normal dental eruption and to avoid any biological damage by probable impaction of two permanent teeth and increase of orthodontic treatment time. This intervention normalised the eruption process (Fig. 2) and the spontaneous eruption of both permanent teeth was obtained (Fig. 3). The patient was re-evaluated at 12y 11m of age and orthodontic therapy was undertaken (Fig. 4, 5, 6) using multibracket appliance according to the Roth prescription (Ovation, Gac Dentsply-Sirona), through a simple sequence of arches and the correction of small orthodontic problems. Optimal orthodontic results were achieved (Fig. 7, 8). Results were found stable at the age of 25, more than 10 years after the end of orthodontic treatment (Fig. 9, 10).

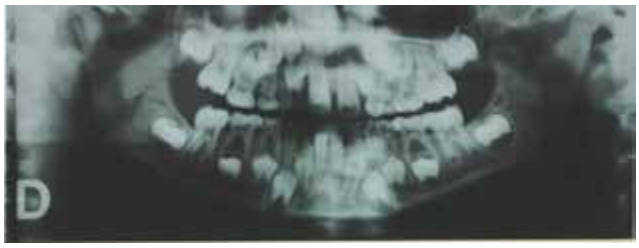

FIG. 1 Compound odontoma in the anterior left mandibular area (Case 1).

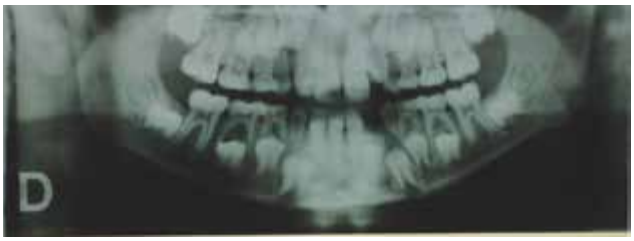

FIG. 2 First radiogram of a x-rays series taken annually to monitor tooth eruption after surgical removal of the odontoma.

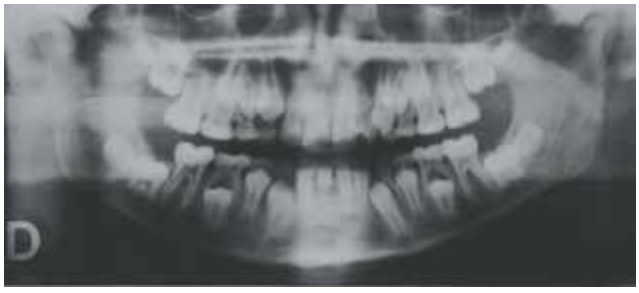

FIG. 3 Panoramic x-ray at the 2-year follow-up showing spontaneous eruption of tooth 3.2 an good positioning of tooth 3.3.

FIG. 4 Pre-treatment lateral x-ray.

FIG. 5 Extraoral images taken before starting orthodontic treatment (12y11m).

FIG. 6 Intraoral images before starting orthodontic treatment (12y 11m), showing spontaneous eruption of teeth 3.2 and 3.3.

FIG. 7 Intraoral images of orthodontic outcome before debracketing

FIG. 8 Lateral x-ray at the end of therapy.

FIG. 9 Extraoral images at the 11-year follow-up.

FIG. 10 Intraoral images at the 11-year follow-up.

Case report 2

A healthy girl came to our observation at the age of 10y 3m for a skeletal and dental Class II malocclusion characterised by a moderate upper and lower crowding. The profile was convex with chin retropositioning and an accentuated labiomental groove (Fig. 11). Intraoral examination showed the lack of eruption of tooth 41 and the permanence of tooth 81 in the dental arch (Fig. 12). Lateral teleradiograph indicated a mesofacial type characterised by reduced SNA and SNB angles (78.2; 76.6) (Fig. 13). An alteration of the eruptive path of tooth 41 was evident due to the presence of a calcified mass in the apical region of tooth 61 (Fig. 14). This condition is also visible in a panoramic radiograph taken by a previous dentist (Fig. 15), showing that in 2 years there had been a considerable alteration of the eruption process of tooth 41 and that, most likely, the early removal of odontoma could have allowed a better or normal eruption of the permanent tooth.

A surgical-orthodontic therapy for the retained ectopic tooth was then initially attempted, using a CBTC in order to better plan the procedure (Fig. 16). Upper and lower fixed appliances where then applied, significantly increasing the torque of the lower incisors with the aim of moving the root tips of tooth 42 and 31 away from the eruptive path of tooth 41, thus creating space for the normal eruption of the retained tooth by a coil spring. Then disinclusion was carried out by a lingual arch with hooks and a vestibular lever starting from a cross tube at tooth 44 (Fig. 17). After two years of treatment, however, due to the eruption in the alveolar mucosa (completely ectopic compared to the natural position) and for biological and timing reasons, in agreement with the patient it was decided to remove tooth 41 and to perform lower spaces closure. This procedure provided for an increased torque of the lower incisors and stripping in the upper arch in order to manage the anterior Bolton index discrepancy and to ensure a correct dental class.

FIG. 11 Extraoral photographs of case 2.

FIG. 12 Intraoral photographs.

FIG. 13 Lateral x-ray.

FIG. 14 Panoramic x-ray shows abnormal position and some structural anomaly of 41 follicle.

Dental compensation in this second phase of the treatment allowed to obtain an overall harmonious profile (even if not greatly improved from the beginning of the therapy, Fig. 18) and an optimal dental occlusion (Fig. 19). Radiographs report the proclination of the lower incisors and a slight retroclination of the upper incisors (Fig. 20, 21). Results remains stable even after 5 years (Fig. 22, 23).

Discussion

Our study aims to underline the important issue regarding the timing and optimal treatment sequence for the correct therapeutic approach to odontoma, to reduce the orthodontic problems that they entail. Odontoma must be evaluated also in the broader context of tooth abnormalities of number

FIG. 15 Panoramic x-ray (taken by a previous dentist 2 years before our first visit) shows the presence of an odontoma causing the retention of tooth 41.

FIG. 16 3D reconstruction from a mandible CBCT.

FIG. 17 Disimpaction mechanics of tooth 41.

FIG. 18 Final extraoral photographs.

FIG. 19 Final intraoral photographs.

FIG. 20 Final panoramic x-ray.

FIG. 21 Final lateral x-ray.

FIG. 22 Extraoral photographs taken at the 5-year follow-up.

FIG. 23 Intraoral photographs taken at the 5-year follow-up.

and position with their particular biomechanical approaches [Paduano et al., 2016, 2014, 2013, 1988], post-treatment stability related to skeletal discrepancies [Aiello et al., 2021] and general condition of oral health in the paediatric population [Paduano et al., 2018]. As shown in the first case, early surgical removal of the odontoma can lead to a normal eruption, reducing or preventing the biomechanical and biological problems of surgical-orthodontic management of retained teeth. A late approach, as shown in the second case, can lead very often to anomalous tooth eruption process or retention, forcing the orthodontist to use complex mechanics and long and expensive therapeutic procedures, thus increasing stress on the patient. Sometimes removal of the retained tooth represents the best therapeutic choice to allow a more conservative approach and avoiding serious problems to the adjacent teeth. Although it is possible to achieve excellent results even in the congenital agenesis of one or two lower incisors [Paduano et al., 2021], it is clear that the most desiderable outcome would be to bring all impacted teeth into correct occlusion. Therefore, the late treatment of an odontoma can compromise the tooth eruption process up to conditions very difficult for the orthodontist to manage.

Conclusion

Early diagnosis, timing, optimal treatment sequence and multidisciplinar approach are the key factors in the treatment of odontomas associated with impacted teeth.

Screening of younger patients by panoramic radiographs together with the accurate examination during primary dentition and early mixed dentition is essential for an efficient interceptive treatment of odontomas, avoiding in many cases the need of orthodontic or surgical-orthodontic treatment, extractions or other challenging interventions such as autotransplantation.

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