CASE REPORT

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Interdisciplinary (surgical and orthodontic) treatment in a patient with compound odontoma associated with impacted permanent lateral incisor of the mandible – a case report

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Keywords

impacted tooth, odontoma, orthodontic treatment

SUMMARY

Impacted teeth are one of the reasons for orthodontic appointments. Two conditions must be met for the tooth to be considered impacted, i.e. a fully developed tooth that has not erupted on time. The presence of an odontoma in the maxillary bone when the impacted teeth are observed is a particular situation. We present a case of a patient with a compound odontoma located in the anterior part of the mandible and associated with an impacted right lateral incisor, who received interdisciplinary (surgical and orthodon-tic) treatment.

The aim of the study was to present the advances in interdisciplinary (surgical and orthodontic) treatment of the compound odontoma associated with the impacted permanent lateral incisor of the mandible and a review of relevant literature.

A literature review covering years 1990-2016 was performed. Medical databases, such as PubMed and Medline, were used. The following keywords were used: "impacted tooth", "odontoma", "orthodontic treatment". Additionally, a manual search of specialist literature on orthodontics and maxillofacial surgery was conducted. Written consent from the subject/subject's parents/subject's legal guardians was obtained. Polish- and English-language publications were included.

Early diagnosis of odontoma enables less complicated and less expensive treatment. The treatment plan should be tailored to each patient. More research is needed on predicting the behaviour of impacted teeth related to odontomas.

INTRODUCTION

Impacted teeth are one of the reasons for orthodontic appointments. Two conditions must be met for the tooth to be considered impacted, i.e. a fully developed tooth that has not erupted on time. A fully impacted tooth is completely embedded in the bone, whereas a partially impacted tooth is covered with mucosa or only a fragment of the tooth's crown is erupted (1). There are a number of reasons for the presence of impacted teeth, such as e.g. lack of space in the dental arch (due to a tendency to reduce mandibular and maxillary bones in the course of evolution as well as inconsistencies in the number and size of teeth), traumatic and genetic factors, inflammatory processes within the bone, vitamin A and D deficiency, and endocrine disorders. Impacted teeth also occur in syndromes, e.g. cleidocranial dysostosis (1, 2). The presence of odontoma in the maxillary bone when impacted teeth are observed is a particular situation. We present a case of a compound odontoma located in the anterior part of the mandible and associated with impacted permanent right lateral incisor of the mandible.

The aim of the study was to present the advances in interdisciplinary (surgical and orthodontic) treatment of a compound odontoma associated with the impacted permanent right lateral incisor of the mandible and a review of relevant literature.

A literature review covering years 1990-2016 was performed. Medical databases, such as PubMed and Medline, were used. The following keywords were used: "impacted", "odontoma", "orthodontic treatment". Additionally, a manual search of specialist literature regarding orthodontics and maxillofacial surgery was conducted. Polish and English-language publications were included.

CASE REPORT

A 17-year-old female patient reported to the Orthodontic Clinic in the Specialist Hospital in Radom for consultation and treatment due to a persistent mandibular right lateral incisor and a bulging on the internal surface of the mandible. No significant family or general medical history was reported. No previous trauma or infections in the anterior mandible were reported. Extraoral examination revealed no abnormalities. Intraoral examination revealed mixed dentition, and in the right mandible: a persistent primary lateral incisor, the lack of permanent lateral incisor and oedema of the inner surface of the anterior mandible (fig. 1a-e). The adjacent teeth (mandibular canine tooth and mandibular canine tooth on the right side) showed normal colour and reaction to ethyl chloride. The surrounding mucous membrane was smooth, pink, moist and shiny. Orthodontic examination showed Angle's Class II tendency and 1/2 of cuspid class II on the right, Angle's Class I and cuspid class I on the left, as well as: a midline shift of 3 mm to the right, minor maxillary and mandibular dental crowding.

Panoramic radiography (fig. 2) showed an oval structure with clear borders and filled with a mass of small structures, probably corresponding to a compound odontoma. Cephalometry showed skeletal class I and anteriorotation (fig. 3). Cone beam computed tomography (CBCT) of the mandible was also preformed to precisely locate the lesion and plan treatment. CBCT showed no resorption of the teeth adjacent to the tumour, i.e. the medial incisor and the canine tooth (fig. 4).

After clinical and radiological examination, a preliminary diagnosis was made: a compound odontoma in the anterior part of the mandible, impacted mandibular right lateral incisor. The treatment plan included interdisciplinary, i.e. surgical and orthodontic, treatment. A two-stage surgical treatment was planned. In the first stage, surgical enucleation of the tumour was performed, while the second stage involved tooth exposure and positioning of a bracket on the tooth in order to apply orthodontic force. The aim of orthodontic treatment was to restore the site for the impacted permanent mandibular lateral incisor, and then introduce the tooth into the dental arch using orthodontic force. Written consent from the subject/subject's parents/subject's legal guardians was obtained.



Fig. 1a-e. Intraoral photographs before treatment showing a persistent deciduous right lateral mandibular incisor



Fig. 2. Panoramic radiograph showing a diagnostic suspicion of an odontoma and impacted permanent mandibular right lateral incisor

∢SNA	78°
∢SNB	77°
∢ANB	1°
∢SNPo	79°
WITS	0mm
∢ML:SN	36°
SGo:NMe%	68
<u>∢1</u> :NS	94°
<u>1</u> :NPo	4mm
<u>∢1</u> :Ī	141°
∢ī:ML	94,5°
ī:APo	0mm

Fig. 3. Cephalometric findings (Steiner modified by Kaminek)

The interdisciplinary (surgical and orthodontic) treatment was initiated by placement of upper and lower light fixed archwires (Elite Opti MIM 022 Roth). A passive spring was used to maintain the site after extraction of persistent primary right lateral mandibular incisor. This was followed by the first stage of surgical treatment, which was performed under general anaesthesia due to the large size of the odontogenic lesion (12.4 mm) and its unfavourable location. A complete removal of



Fig. 4. CBCT showing the position of the lesion and the impacted permanent mandibular right lateral incisor

mandibular tumour and persistent tooth was performed. Histopathological examination confirmed the primary diagnosis of compound odontoma. A steel wire 0.018 inch in diameter was placed on the lower dental arch after 3 months. The second stage of surgical treatment, i.e. impacted tooth exposure and placement of an orthodontic bracket with the introduction of metal ligatures, was performed (fig. 5). This was followed by metal ligature activation sessions during subsequent visits. Five months later, impacted tooth re-exposure was necessary due to orthodontic bracket detachment.

Three x-ray scans targeting the region of the right lateral mandibular incisor were performed during orthodontic treatment. The first image (at month 12 after treatment onset) showed an improvement in the position of the impacted tooth and the need for a change in the direction of activation. The two other images (months 17 and 22 after treatment onset) showed progressing eruption



Fig. 5. Intraoral photograph showing the impacted permanent mandibular right lateral incisor with the orthodontic bracket

of the impacted tooth (fig. 6a-c). At present, orthodontic guidance of the impacted tooth into the dental arch is continued.

DISCUSSION

Odontomas are the most common odontogenic tumours (20-67% of all lesions of this type) (3, 4). Since they contain epithelial and mesenchymal component (5, 6), they are classified as mixed odontogenic tumours. The World Health Organization (WHO) classifies odontomas into two types (7, 8):

- complex odontoma (lat. odontoma complexum)
 chaotically arranged mass of dentin, cementum and enamel,
- compound odontoma (lat. odontoma compositum)
 a cluster of odontoids, i.e. structures composed of enamel, cementum, dentin and pulp, with normal arrangement resembling small teeth (3, 9, 10).

Additionally, the following odontomas may be encountered in the clinical practice: mixed odontoma (a tumour composed of complex and compound odontoma) (11) and erupted odontoma (currently considered to be a compound odontoma which erupted into the oral cavity). Only limited cases of odontomas which erupted into the oral cavity have been reported in the literature (3, 9, 12-16). According to the WHO and the International Agency for Research on Cancer (IARC), odontomas are hamartomatous rather than neoplastic lesions (7, 17, 18). They may occur in the case of familial colorectal polyposis (Gardner's syndrome) (19), Herman syndrome (17, 18, 20), basal cell nevus syndrome and Tangier disease (9). The etiopathogenesis of odontomas in unknown. Traumatic, infectious and genetic aetiology is suspected (3, 10). Complex odontomas account for 30% of odontogenic tumours. They are usually diagnosed in the second decade of life (17). They are much more common in men (men: 50.6%, women: 49.4%) (21). According to various sources, compound odontomas account for 47-74% of odontogenic tumours (17, 18, 22). They

are diagnosed earlier than complex odontomas (i.e. 75% of cases are diagnosed before the age of 20 years) (17, 21). Odontomas are most often associated with permanent dentition (23, 24). Mixed odontomas are usually located in the lateral sector of the mandible (25), while compound odontomas are most often found in the anterior sector of the maxilla (9, 26). In the presented case, the location of the compound odontoma was not typical, i.e. in the anterior sector of the mandible.

Clinically, odontomas are painless and slow growing. In most cases, they are diagnosed accidentally during an X-ray, with impaired tooth eruption being their first symptom (10-40%) (4, 27). Other symptoms include dislocation of adjacent teeth, alveolar distension (11, 17, 18, 28, 29), very rarely pain and inflammation (3). Our patient reported to the Specialist Hospital in Radom due to persistent primary right lateral mandibular incisor and a minor bulging in this area. The radiological appearance of complex odontoma depends on its stage of development. An earlystage odontoma may appear non-typically radiolucent, then as a radiolucent-radiopaque structure, and finally as a radiopaque lesion with amorphous masses of dental hard tissue surrounded by a thin radiolucent zone (30, 31).

Radiologically, a compound odontoma appears pathognomonic: a round or oval lesion surrounded by a radiolucent zone and filled with small tooth-like structures (32). Resorption of teeth adjacent to the tumour is very rare (4). Odontomas should be differentiated from ameloblastic fibroma, ameloblastic fibro-odontoma, and ameloblastic odontoma (21, 33).

Complete excision of odontoma is the treatment of choice. Histopathological examination of the excised tumour is necessary (4, 16, 34). The coexisting impacted tooth may be preserved if it does not affect surgical radicality, can be introduced into the dental arch and shows no signs of resorption (4). It is still disputable whether an impacted tooth located in the immediate vicinity of the tumour or a resorbed tooth may be preserved (35).



Fig. 6a-c. Periapical radiographs showing position improvement of the impacted tooth during orthodontic treatment

The time of procedure after the diagnosis of odontoma depends on the child's age. According to some of researchers, the treatment should be postponed until the roots of permanent teeth adjacent to the lesion reach at least half of their length, if possible (24). Kaban (36), on the other hand, claims that the risk of damage to the teeth adjacent to the tumour is low provided that the procedure is performed carefully, and the teeth are separated with a bony septum from the odontoma. Morning recommends early excision of an odontoma (37). According to Ashkenazi et al., the chances of spontaneous eruption of an impacted tooth decrease by 2% with each month of delay (38).

The prognosis for odontomas is considered to be very good (7). Recurrence is very rare and is most often observed in non-mineralised odontomas (9, 39, 40). Clinical and radiological follow-up is necessary (34). According to An et al., the chances of retaining impacted teeth decrease with patient's age. This relationship is statistically significant (p < 0.01). In the group of younger patients (below 9 years of age), 100% of impacted teeth were preserved, whereas all impacted teeth associated with odontomas were removed in patients over 30 years of age (35). Common malocclusions associated with odontomas require orthodontic treatment (17).

Two types of management may be implemented to preserve an impacted tooth associated with odontomas: allowing spontaneous eruption (with or without impacted tooth exposure) or applying orthodontic forces to introduce the tooth onto the dental arch already during odontoma enucleation. The chances for spontaneous eruption of an impacted tooth range between 32% (Tomizawa et al., Ashkenazi et al.) and 45% (Morning) (37, 38, 40). Researchers suggest different criteria that should be taken into account when planning a procedure for an impacted tooth associated with odontoma. Some authors indicate that the chances for spontaneous eruption of an impacted tooth increase if the tumour is small, impacted tooth position and shape do not deviate significantly from normal, the impacted tooth has incomplete root development, and there is sufficient space in the dental arch (41, 42). Hisatomi et al. indicate that the planning of management in an impacted tooth depends on the level of development of the impacted tooth at the time of odontoma excision. However, an impacted tooth may erupt in the wrong position (43). Many authors point out that complete root development is an indication for the use of orthodontic forces during surgical tumour removal (41, 44-46).

Ashkenazi et al., showed in their study that the following factors may be important for the prognosis of spontaneous eruption of an impacted tooth: (1) the angle between the long axis of an impacted tooth and the palatal suture line, (2) displacement of the apex of the impacted tooth. However, the authors excluded the correlation between the chance for spontaneous eruption of an impacted tooth and the level of development of the impacted tooth. Ashkenazi et al. also showed that spontaneous eruption of an impacted tooth is also affected by the shape of the obstacle. Supernumerary teeth with irregular shape and odontomas reduce the chances for spontaneous eruption of an impacted tooth. The degree of spontaneous eruption of impacted teeth associated with irregularly shaped impacted tooth and associated with an odontoma was 85% lower compared to impacted teeth associated with a cone-shaped structure, normal shape or reduced-size supernumerary teeth (38).

It is still discussed by researchers whether orthodontic forces used to guide an impacted tooth to the dental arch should be applied during odontoma enucleation or later. Tomizawa et al. conducted their study in a group of children aged between 1.2 years and 14 years. The authors suggested a 3-month observation period after tumour removal for spontaneous eruption of an impacted tooth (40). latrou et al. and Ashkenazi et al., on the other hand, propose that orthodontic forces be used simultaneously with odontoma enucleation to avoid another surgery (45).

In our case, two-stage surgical treatment was implemented due to the large size and the shape of odontoma. The second stage involved impacted tooth exposure and bracket placement due to unfavourable prognosis for spontaneous eruption (patient's age, completed root development, curved root apex, impacted tooth dislocation). The final X-ray showed no odontoma recurrence and significant improvement in the position of the impacted tooth.

CONCLUSIONS

Early diagnosis of odontomas allows for a less complicated and less expensive treatment (24, 38, 47). The recommendation of Kodali et al. on routine panoramic radiography aimed at early detection of this odontogenic benign tumour seems reasonable (48). An et al. suggest that panoramic radiography should be performed in the first and second decade of life (35). Treatment plan should be tailored to each patient so as to maintain all impacted teeth, if possible. Further studies to investigate the prognosis for the preservation of impacted teeth associated with an odontoma are needed.

CONFLICT OF INTEREST

None

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