

Giant Complex Odontoma of the Mandible: A case report and literature review

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Abstract

Odontomas constitute a developmental defect of hard dental tissues and are classified as benign odontogenic tumours. They are composed of all dental structures and tissues: enamel, dentin, cementum and pulp. As regards histomorphological features, two types of odontomas have been differentiated: complex and compound. Odontomas represent from 4.7% to 76% of odontogenic tumours. Their aetiology has not been fully understood. Injury, infection and genetic factors are often named among the causes. Odontomas are considered a type of odontogenic hamartoma, and are generally reported not to exceed 3 cm in diameter. As hamartomas, giant odontomas generally show no signs or symptoms, and are usually detected by chance in radiographic images taken in relation to disrupted eruption or mislocation of teeth, or if they perforate the mucosa to become exposed in the oral cavity. To present the case study of an erupting complex odontoma on the left side of the mandible in an 11-year-old girl. The analysis covered medical documentation of the patient, diagnostic casts, orthopantomographs and description of operative protocol. The case study has been complemented with the review of up-to-date literature. A lesion composed of osteoid tissue was removed during one-day surgery without subsequent augmentation with a bone substitute material. There were no post-surgery complications. A follow-up orthopantomograph taken 2 months and 2 years later showed that the wound had been healing correctly. Odontomas are benign lesions that can be removed during one-day surgery without the absolute need for augmentation with xenogenic or allogenic material.

Keywords: complex odontoma, clinical manifestations, radiographic examination.

1. Introduction

Odontomas are a developmental defect of hard dental tissues. They have been classified by the World Health Organisation (WHO) as benign, tumour-like odontogenic lesions (hamartoma) and are generally reported not to exceed 3 cm in diameter. They are located in the maxillofacial area, in bones and adjacent tissues (Barnes L *et al*, 2005) and are composed of all of the dental structures and tissues, i.e. enamel, dentin, cementum and pulp (Tyagi P *et al*, 2010). Odontomas can be differentiated according to their anatomical presentations: Compound odontoma - clusters of several denticles and complex odontoma - well defined tumefaction mass. The diagnosis can be performed by radiographic examination. Odontomas are further sub-classified based upon their gross and radiographic features into compound (small tooth-like structures), complex (a conglomeration of dentin, enamel and cementum) and cystic (Neville BW *et al*, 2009).

The aetiology of odontoma formation has not been fully understood. They develop from epithelial and mesenchymal components of the dental apparatus, producing enamel and dentin. They can occur at any age, but are most common in the first two decades of life, with an average age of 14–18 years. Other probable factors include genetic issues such as odontoblastic hyperactivity and alterations in the genetic components responsible for controlling dental development, Gardner syndrome, or Hermann syndrome. Odontomas are generally small; however, they may occasionally grow large, resulting in the bone expansion (Oz YG *et al*, 2007).

Histologically, compound odontomas have the tooth-like structures which are arranged in a uniform manner similar to the normal tooth. The structures in complex odontomas are mixed and disorganized (Srivastava A *et al*, 2012).

Differential diagnosis considers: Ameloblastoma, myxoma, ameloblastic fibro odontoma, cementoblastoma, ameloblastic fibroma, and calcifying epithelial odontogenous tumor which were more frequently located in the posterior region of the mandible (Guerrisi M *et al*, 2007).

A recurrence is not described; if it recurs, one must rule out other odontogenic lesions such as calcifying odontogenic cyst and ameloblastic fibro-odontoma (Reichart PA *et al*, 2004; Chen Y *et al*, 2005).

Conservative approach by means of surgical enucleation or curettage is the treatment of choice. Giansanti *et al*. reported after he followed a number of cases from 1 to 10 years, showed the absence of recurrence after local curettage of the tumor (Giansanti *et al*, 1970).

In general recurrence rate of Odontoid Tumors is rare, but only a few cases from Japan were noted (Reichart PA *et al*, 2004).

The aim of this article is to present an own case study of a complex odontoma in an 11-year-old female patient. We have applied the WHO classification and reviewed the up-to-date literature on the subject.

2. Case study

The patient, A.S , aged 11, reported to the University Clinic for maxillofacial surgery with atypical dental pain, reporting an excessive pressure and pain sensation in the area of the left first lower molar (36). Clinical examination did not indicate any signs of inflammation. In addition, there was a free edentulous space beyond the second premolar (35) and the first molar was missing. The patient was referred for radiographic examination (orthopantomographs), which showed an irregular, radio-opaque tooth-like structure in the area of teeth 36 and 37 (Figure 1).

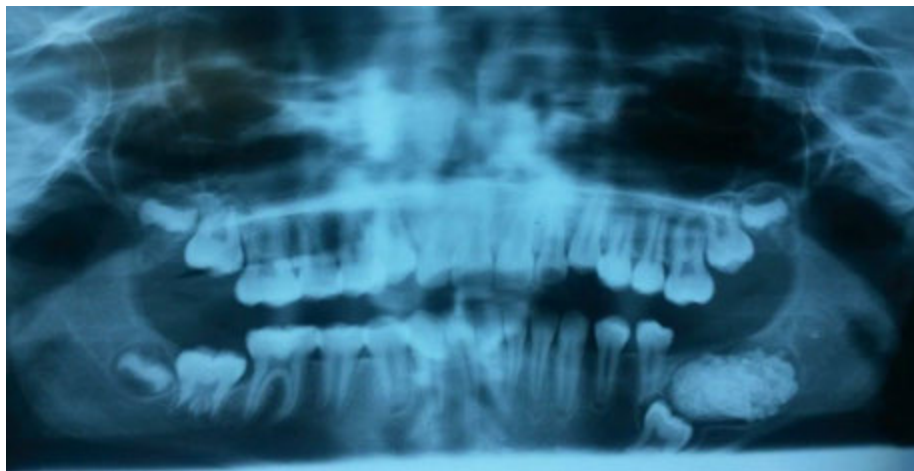


Figure 1. Complex odontoma of left side of the mandible with impacted 36

The clinical and X-ray examination revealed a highly mineralised tumour **42mm x 28mm**, composed of osteoid tissue with radio-opacity similar to the tooth structures and allowed made it possible to diagnose a complex odontoma in the mandible. After consulting and informing the parents the decision was taken to remove the lesion during the one –day surgery. The procedure was performed in general anaesthesia. Once the mucoperiosteal flap had been detached, a fragment of the bone overlaying the odontoma was removed from the vestibular side. Then, the lesion composed of hard osteoid tissue was excised leaving a relatively big cavity (Figure 2).

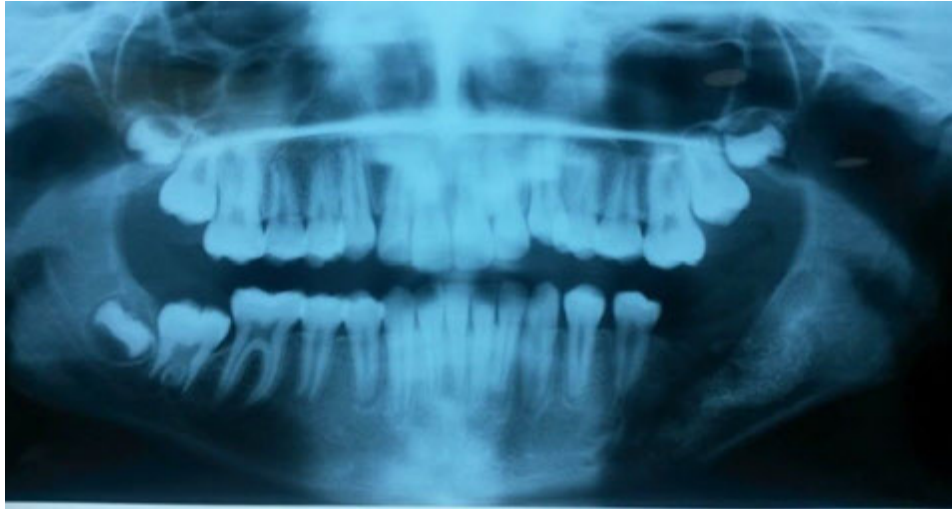


Figure 2. X-ray 2 months after surgery

We did not find a well-defined capsule, but nevertheless, the lesion was well-circumscribed from the surrounding healthy bone.

The impacted first molar (placed almost near the inferior margin of the mandible) was also operatively extracted. Meticulous hemostasis was performed and the wound was closed with sutures without filling the cavity with any bone substitute. The material was sent for histopathological examination which confirmed the diagnosis of a complex odontoma (Figure 3).

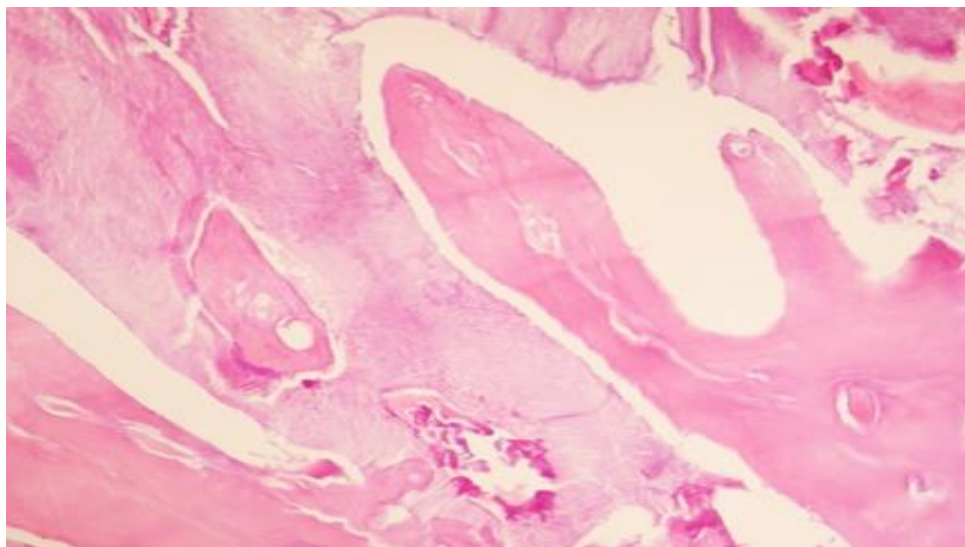


Figure 3. Histopathology of Complex Odontoma - disorganized mass of randomly-arranged dentin intermixed with enamel matrix

After 10 days the wound was declared to heal correctly and the sutures were removed. Follow-up orthopantomographs taken two months later (Figure 2), confirmed that the wound healing process was correct. Two years later, orthopantomograph showed complete healing and mineralization of the space of the operated area as well as the follicle of the third molar was visible (Figure 4).



Figure 4. Two years after. Notice the mineralization and the follicle of 38

3. Discussion

In our case, the histopathological examination indicated an odontogenic tumor and, combined with the clinical and intraoperative picture, confirmed the diagnosis of a complex odontoma. Due to the lack of distinct pain symptoms (not in our case), odontoma-type lesions are usually discovered by chance – in radiographic images taken because of disrupted eruption or dental anomalies in malocclusion (Khan N, Shrivastava *et al*, 2014).

In radiographic images, lesions are visible as a well-defined radio-opaque focus surrounded with a radiolucent halo or as a number of such foci representing odontoids (in compound odontoma). Their presence may be indicated by bone expansion, slight expanding pain, delayed or arrested eruption of permanent teeth or persistent deciduous teeth (Venigalla A *et al*, 2015). Compound odontomas are more common in the anterior jaws, while complex odontomas occur more often in the posterior jaws.

Rarely they can be placed in the soft tissues as buccal mucosa reported by Venigalla *et al*, 2008.



Figure 5. Intraoral view showing eruption of tooth-like mass from right buccal mucosa

In our case, we did not identify persistent deciduous teeth. There was missing permanent teeth in the mandible and a free toothless space posterior to the second premolar of the left side.

Compound odontomas are usually composed of several odontoids and their size oscillates (Lee ChH *et al*, 2008). A similar number of odontoids (37 specimens) was identified by Kubasiewicz-Ros *et al*, 2015 and by Niharika *et al*, 2015 at a different location (the anterior section of the maxilla).

In the case described by Jankowska *et al*, a complex odontoma removed in a teenager was composed of 19 odontoids (Janowska K *et al*, 2015).



Figure 6. Clinical photograph showing fragments of a complex odontoma with a left mandibular impacted third molar

Odontomas are diagnosed usually in the 2nd decade of life (63%), less often in the 3rd one (24%) (Khan M *et al*, 2011). They are usually associated with permanent dentition and appear sporadically in deciduous dentition (Mathew AK *et al*, 2013). The analysis of odontoma cases treated in the Dental Surgery Institute of the Medical University of Łódź (Zakład Chirurgii Stomatologicznej UM w Łodzi) in 1990-2004 indicates that this type of lesion affects more often men than women, at the ratio of 7:2. The research conducted by Janas (2008), confirmed that, in relation to the earlier studies, complex odontomas occur more often in men than women, at the ration of 11:7, yet they were more often located in the mandible.

An extensive meta-analysis based on 30 academic publications containing 3065 cases of odontoma demonstrated that out of 1761 cases of odontoma 49.4% were represented by women and 50.6% by men. Moreover, out of 1340 cases, 61.3% were identified as compound odontoma, 37% as complex odontoma and 1.7% were not classified as either (Hidalgo-Sánchez O *et al*, 2008).

The treatment consists in complete removal of the tumor. As the lesion is encapsulated, it is possible to dissect the odontoma from the surrounding bone during the surgery. Similarly, also in the case of our patient, the odontoma located in the mandible was fully removed.

Some authors fill the cavity created after the removal of the odontoma. Opinions on that matter vary – some of the authors (Kiewlicz W. *et al*) report that bone deficiency was complemented with Cerasorb (Curasan) and secured with a resorbable Bio-Gide membrane, others close the cavity tightly with sutures. We closed

the cavity without filling it with bone substitute material. The follow-up orthopantomograph showed that the healing process was correct.

In the presented case, we did not determine the aetiological factor, which might have contributed to the complex odontoma. A possible cause might have been a persistent infection in early childhood and the resulting later long-term antibiotics therapy, whereas its manifestation and eruption at the age of 11 might have been triggered by the age of the patient.

4. Conclusions

Odontomas are benign lesions that may be excised during a one-day surgery. Giant complex odontoma is a very rare entity. The pressure exerted by the plate of a removable appliance during the orthodontic treatment may initiate the eruption of the odontoma. Additionally, cases of giant odontomas can be distinguished from other conditions mostly by the age of the affected patient, the position of the lesion, its sex predilection, and radiological features including differences in the opacity of the radiopaque mass and features of the well-defined radiolucent rim. In young patients a slight bone deficiency is left after the excision of an odontogenic tumor heals correctly without the need for augmentation with bone substitute material (as this is up to the individual decision of the surgeon).

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