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Compound odontoma in a 9-year-old male patient – Case report

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CASE STUDY

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ABSTRACT

Odontomas are odontogenic benign tumours composed of hard dental tissue formed from the epithelial and mesenchymal base. Although odontomas are sometimes suspected in case of delayed eruption of a permanent heir or a persistent deciduous tooth in the dental arch, they are usually asymptomatic and are incidentally found during routine radiographic imaging. Odontomas are histopathologically divided into complex and compound odontomas. We here describe a 9-year-old male patient with compound odontoma, thereby wish to summarize this tumour: its diagnosis and treatment.

Key Words
Benign odontogenic tumours, mixed odontogenic tumours, compound odontoma

Implications for Practice:

1. What is known about this subject?
Compound odontoma generally occurs as a painless formation in frontal maxillar region. The condition is often associated with prolonged persistence of deciduous teeth.

2. What new information is offered in this case study?
This study shows a rare case of retained permanent teeth associated with odontoma and it may be of some clinical use in treating a paediatric patient with this tumour.

3. What are the implications for research, policy, or practice?
Patients presenting with compound odontoma should undergo surgical removal of calcified tissues and enucleation of the connective sheath.

Background

Odontomas are the most common benign mixed odontogenic tumours consisting of fully differentiated cells of epithelial and mesenchymal origin.[1,2] Due to their composition and non-aggressive nature, odontomas are often called hamartomas.[3]

Odontomas can occur at any age, although a slightly higher prevalence has been observed in children and adolescents.[4] They occur equally in both sexes. The aetiology is still not completely clear, but it is assumed that such formations are associated with odontoblastic hyperactivity.[5,6]

Histological analysis of odontoma shows the presence of enamel matrix, dentin, cementum and pulp tissue, which may or may not achieve a normal degree of morphodifferentiation.[7] According to histopathological features, the WHO classification differentiates odontomas as complex and compound odontomas. Complex odontomas are neoplasms that are irregular in structure and less differentiated from normal dental tissues. Such unique mass of calcified bone tissue is separated by a connective capsule identical to the follicle surrounding a normal tooth.[8] Complex odontomas most commonly occur in the mandible in the area of the lateral teeth.[9,10] Compound odontomas consist of many small tooth-like structures with pulp-like tissue in the centre, surrounded by dentin and partially covered with enamel.[8] These structures are separated from each other by connective tissue that also separates them from the surrounding bone.

Odontomas generally occur as asymptomatic, painless, slow-growing formations.[11]

Prolonged persistence of deciduous teeth and delayed eruption of permanent teeth often appear as the first clinical signs of odontoma. The most common localization of teeth impacted with compound odontoma are the anterior parts of the maxilla.[1,12] Slightly less frequently and if large enough, odontomas can lead to swelling of the affected area and dilation of the cortical bone which can cause minor facial asymmetries.[3,13,14] Although odontomas most commonly appear as intraosseous lesions, sometimes after full calcification they can erupt like teeth into the oral cavity. Odontoma growth ends with calcification of hard dental tissues.

Odontoma therapy is mostly conservative and involves the surgical removal of hard tooth tissue with enucleation of the connective tissue. The prognosis is extremely favourable.[11,12]

Case details

A nine-year-old boy, accompanied by his parents, came to the Department of Paediatric and Preventive Dentistry at the Faculty of Dentistry, University of Zagreb, with the main complaint of still unerupted teeth in the anterior maxillary region. Medical history does not indicate possible predisposing factors, which could in any way stimulate the formation of odontomas. No diseases or allergies are reported. Also, the patient does not report any trauma to the orofacial region. There is no anamnestic data on familial anodontia, hypodontia or any type of tumour of odontogenic origin.

Clinical intraoral examination palpates a hard spherical formation that protrudes into the vestibule in the area of teeth 51, 52 and 53. The right maxillary deciduous central and lateral incisors are still present, while contralateral teeth have already erupted and are properly positioned in the dental arch (Figure 1). Such asynchronous growth and eruption of teeth, including the palpable formation in the vestibule, indicated the need for orthopantomogram.

Review of the orthopantomogram in the region of teeth 51, 52, and 53 revealed multiple radiopaque lesions of a joint diameter of about 2.5cm within a thin radiolucent fibrous capsule. Teeth 11, 12, and 13 were also observed in the image (Figure 2).

The boy was then sent to the Department of Oral Surgery where, based on the orthopantomogram and clinical findings, a diagnosis of a compound odontoma in region 51 to 53 with retention of permanent heir teeth was made. Based on the diagnosis, surgical extraction of a compound odontoma under local anaesthesia is planned.

Intraoperatively, after removal of the mucoperiosteal flap according to Nowak-Peter and removal of the bone that covered the labial surface of the lesion with a surgical drill, the envelope of the formation was attached. Pieces of hard dental tissue were extracted and the connective sheath was enucleated (Figures 3, 4). Teeth 51 and 52 were also extracted (Figure 5). The flap was then positioned and sutured with a non-resorptive polyfilament silk suture 4/0 (Figure 6).

One week after the operation, the patient came for a check-up. Sutures have been removed and a neat postoperative course was visible (Figure 7). The patient was advised to consult further with an orthodontist.

Discussion

The term odontoma was first used by Paul Broca in 1867 to describe all odontogenic tumours. Today, the term refers only to mixed odontogenic benign tumours, composed of fully differentiated cells of epithelial and mesenchymal origin.[3] Compound odontomas are benign odontogenic tumours with a high degree of differentiation of hard dental tissues whose growth and development ends when the calcification of the tooth is fully completed.[13,15] They are relatively common and histologically they are more often classified as hamartomas than as true neoplasms.[12]

Although they can be found at any age, Siriwardena et al. state in their 2018 study that the average age of odontoma occurrence is 18±13.5 years, with a slightly higher incidence in the male population (M:F, 1.7:1). No significant difference was found between compound and complex odontomas taking into account age and sex, but the largest difference was noted in their localization. Complex odontomas occur more frequently in the posterior parts of the mandible (63%) while in most cases compound odontomas appear in the anterior parts of the maxilla (82%).[16]

Kaemmerer et al.[17] as well as Seo-Young et al.[15] emphasize that most odontomas do not show any clinical signs or symptoms in patients and for the most part (75.3%) are detected as chance orthopantomogram findings. The irregular mass of calcified, radiopaque tissue surrounded by a thin radiolucent halo is suggestive of a complex odontoma. Compound odontoma shows numerous calcified tooth-like structures at the centre of a sharply demarcated radiolucent lesion. Differential diagnosis should exclude ameloblastic fibroma, ameloblastic fibrodentinoma and calcifying epithelial odontogenic tumour (Pindborg

tumour).[11] This data contradicts some older studies in which delayed tooth eruption was the main reason for reporting to the dentist. The most likely reason for this is the fact that in recent times panoramic imaging is more frequently used for routine inspections. On the other hand, if odontomas go unnoticed, sometimes they can remain in the bone without any clinical manifestations.[18] Hidalgo-Sanchez et al. conclude in a meta-analysis that 57% of odontomas cause symptoms such as delayed tooth eruption, swelling, pain, inflammation or infection. In some cases, odontomas can cause tooth spacing and devitalization and rarely root resorption of adjacent teeth.[11,19] According to a retrospective study by Seo-Young et al.[15] conducted on 75 cases, impaction of permanent teeth is the most common symptom (61.6%) by frequency, followed by prolonged retention of deciduous teeth (32.9%) and dislocation of adjacent teeth (30.1%).

Odontomas must be surgically removed to prevent possible cyst formation or possible conversion to odontoameloblastoma.[20] Likewise, ameloblastic fibrodentinoma and ameloblastic fibroma have a great resemblance to the usual odontoma, especially on the radiograph, and it is suggested to do a histological analysis of odontoma before making final diagnosis.[21]

Conclusion

In the presented case, the compound odontoma is presented as multiple calcified tooth-like structures of different size and shape and it contains all hard dental tissue. Odontoma is located in the right anterior part of the maxilla in the region of teeth 51, 52 and 53 which according to numerous studies is the most commonly affected area.[9,15,17] Likewise, odontoma appeared as an asymptomatic and painless formation and due to its size caused retention of permanent central and lateral incisors, as well as permanent right canine. Having in mind all characteristics, including age, sex, localization and symptoms, the presented case mirrors and confirms above referenced studies. As such, this tumour shows some heterogeneous clinical course. In this case, the present surgical procedures succeeded. Further discussion and case accumulation may be needed to conclude how to treat the patients with this tumor and when to do so. The present description may be of some clinical use in treating a pediatric patient with this tumor. Since the surgery was done four months ago, we do not have long-term follow up. Because of that limitation we will inform you about follow up.

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CONFLICTS OF INTEREST

The authors declare that they have no competing interests.

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PATIENT CONSENT

The authors, *Katanec T, Kuterovac L, Katanec D*, declare that:

1. They have obtained written, informed consent for the publication of the details relating to the patient(s) in this report.
2. All possible steps have been taken to safeguard the identity of the patient(s).
3. This submission is compliant with the requirements of local research ethics committees.

Figure 1: Preoperative intraoral examination. Visible spherical protrusion in the vestibule of the teeth in region 51, 52 and 53

Figure 2: Calcified structures in the region of teeth 51, 52 and 53 observed on the preoperative orthopantomogram

Figure 3: Intraoperative presentation of compound odontoma after mucoperiosteal flap elevation

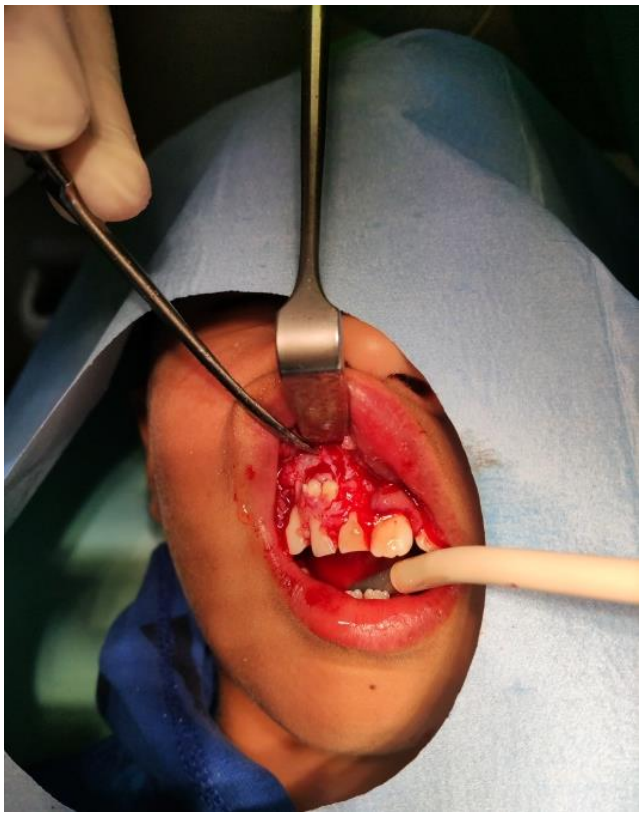

Figure 4: Calcified accumulations of hard dental tissue extracted from the connective sheath of compound odontoma

Figure 5: The resulting bone defect after surgical removal of a compound odontoma and extraction of teeth 51 and 52

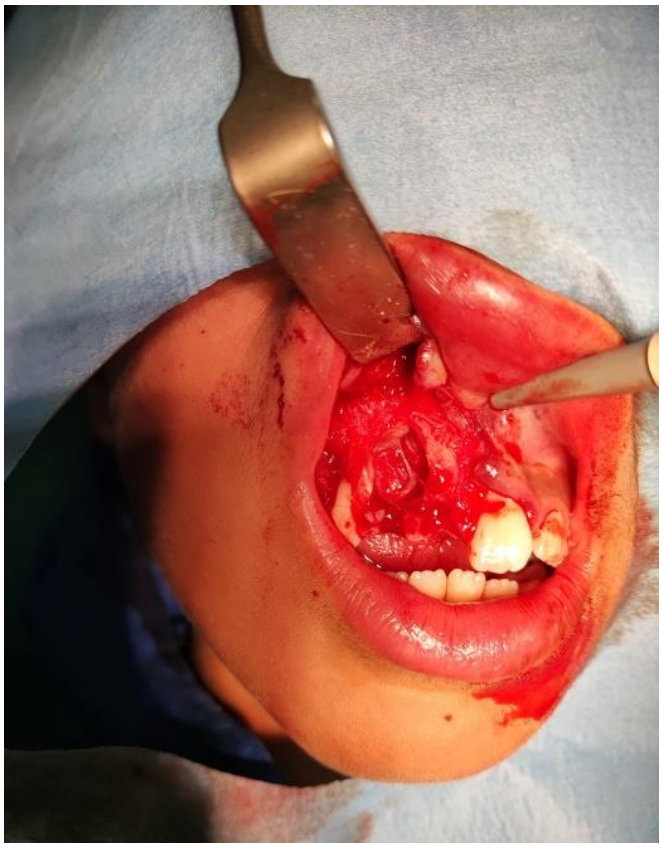

Figure 6: Installed non-resorptive polyfilament silk suture 4/0

Figure 7: Suture removal one week postoperatively