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## COMPOUND ODONTOMA IN THE MANDIBLE – CASE STUDY AND LITERATURE REVIEW

### ZĘBIAK ZESTAWNY UMIEJSCOWIONY W ŻUCHWIE – OPIS PRZYPADKU I PRZEGLĄD PIŚMIENNICTWA

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#### Abstract

**Introduction:** 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, yet injury, infection and genetic factors are often named among the causes. Odontomas are usually detected by chance in radiographic images taken in relation to disrupted eruption or mislocation of teeth.

**Aim of study:** To present the case study of an erupting compound odontoma on the right side of the mandible in an 11-year-old girl.

**Material, methods and results:** The analysis covered medical documentation of the patient, diagnostic casts, orthopantomographs and cone beam computed tomography scans. The case study has been complemented with the review of up-to-date literature.

A lesion composed of 30 odontoids was removed during one-day surgery without subsequent augmentation with bone substitute material. There were no post-surgery complications. A follow-up orthopantomograph taken 3 months later showed that the wound had been healing correctly, which made it possible to plan subsequent orthodontic treatment with a fixed appliance.

**Conclusions:** 1. Odontomas are benign lesions which can be removed during one-day surgery without the absolute need of augmentation with xenogenic or allogenic material. 2. The pressure exerted by the plate of a removable appliance is very likely to initiate the eruption of the odontoma.

**Key words:** compound odontoma, clinical manifestations, radiographic examination – cone beam computed tomography (CBCT), child, orthodontic treatment

#### Streszczenie

**Wstęp:** Zębiaki stanowią wadę rozwojową twardych tkanek zęba. Są klasyfikowane jako łagodne nowotworopodobne guzy zębopochodne. Składają się ze wszystkich struktur i tkanek zęba: szkliwa, zębiny, cementu korzeniowego oraz miazgi. Na podstawie różnic histomorfologicznych wydzielono dwa rodzaje zębiaków: złożone oraz zestawne.

Stanowią od 4,7-76% guzów zębopochodnych. Etiologia nie jest w pełni poznana, a wśród przyczyn wymienia się skutki urazu lub infekcji oraz czynniki genetyczne. Wykrywane są najczęściej przypadkowo na zdjęciach radiologicznych wykonanych z powodu zaburzeń wyrzynania lub położenia zębów.

**Cel pracy:** Przedstawienie opisu przypadku wyrzynającego się zębiaka zestawnego umiejscowionego po stronie prawej żuchwy, u 11-letniej pacjentki.

**Materiał, metody i wyniki:** Analizie poddano dokumentację medyczną pacjentki, modele diagnostyczne, zdjęcia pantomograficzne oraz badanie CBCT. Opis przypadku uzupełniono przeglądem aktualnego piśmiennictwa.

Zmianę składającą się z 30 odontoidów usunięto w ramach chirurgii jednego dnia bez następowej augmentacji materiałem kośćozastępczym. Nie stwierdzono powikłań pozabiegowych. Po 3 miesiącach, kontrolne zdjęcie pantomograficzne wykazało prawidłowe gojenie rany, co pozwoliło zaplnować następowe leczenie ortodontyczne aparatem stałym.

**Wnioski:** 1. Zębaki są zmianami łagodnymi, które mogą być usunięte w ramach chirurgii jednego dnia bez bezwzględnej konieczności augmentacji materiałem kseno- lub allogennym. 2. Ucisk płyty ortodontycznego aparatu zdejmowanego może z dużym prawdopodobieństwem przyczynić się do wyrzynania zębiaka.

**Słowa kluczowe:** zębiak zestawny (odontoma compound), objawy kliniczne, badanie radiologiczne – tomografia komputerowa z promieniem stożkowym (CBCT), dziecko, terapia ortodontyczna

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## INTRODUCTION

Odontomas are a developmental defect of hard dental tissues. They have been classified by the World Health Organisation and International Agency for Research on Cancer (IARC) as benign, tumour-like odontogenic lesions (hamartoma). They are located solely in the maxillofacial area, in bones and adjacent soft tissues [1] and are composed of all of the dental structures and tissues, i.e. enamel, dentin, cementum and pulp [2].

The aetiology of odontoma formation has not been fully understood. Potential causes list inflammation or chronic injury experienced during odontogenesis. Other probable factors include genetic issues such as odontoblastic hyperreactivity, Gardner syndrome or Hermann syndrome. Despite the fact that odontomas are odontogenic tumours, the mechanism of their formation is different from the mechanism of tooth formation, as there is no periodontal ligament around the root [3].

The literature specifies two types of odontoma classification.

The WHO classification from of 2005 recognises odontomas as a developmental disorder. They have been defined as a disorderly mixture of odontogenic epithelium and ectomesenchyma. The above classification distinguishes: compound odontomas – dental tissues are arranged correctly and their appearance resembles small “toothlets” called odontoids. In such a case, dental tissues may be morphologically incorrect, however they are arranged in the same manner as in a correctly formed tooth [1].

Another division, based on the histomorphological features, also differentiates two types of odontomas: composite and mixed. Composite odontomas (odontoma compositum) are characterised by numerous toothlets resembling correct teeth in shape. Those malformations, called odontoids, are composed of morphologically

mature enamel, dentin and cementum. Mixed odontomas (odontoma mixtum) are composed of all dental tissues arranged in a disorderly manner [4].

The studies on odontogenic tumours show that the prevalence of odontomas is diversified, i.e. in the USA, out of 1088 odontogenic tumours, 826 were identified as odontomas (76%) [5], whereas an extensive research conducted in China showed that odontomas represented 4.7% of the 1642 analysed cases of odontogenic tumour-like lesions analysed (complex: 3.5%, compound: 1.2%) [6].

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

## CASE STUDY

The patient, J.B., aged 11, reported to the Orthodontic Clinic at Wojskowa Specjalistyczna Przychodnia Lekarska (sSpecialised mMilitary Ooutpatient mMedical cClinic) with dental anomalies and diastema. The orthodontic therapy was initiated with removable appliances, i.e. an upper and lower Schwarz plate.

During the active phase of the therapy, the patient reported excessive pressure of the appliance and pain sensation in the area of the lower right canine (43) – the tooth showed signs of distorotation (fig. 1). Clinical examination showed the presence of a hard elevation on the lingual side with an erupting mineralised formation on the top. The mucous membrane over the elevation did not indicate any signs of inflammation. In addition, there was a gap between teeth 43 and 44. The patient was referred for radiographic examination (orthopantomographs), which showed a set of irregular, radio-opaque tooth-like structures in the area of teeth 43 and 44 (fig. 7). CBCT scans were recommended in order to obtain a more precise diagnosis.



Fig. 1. Odontoma erupting from the lingual side of the alveolar ridge in the mandible (patient J.B.).

Ryc. 1. Widok wyrzynającego się zębiaka od strony językowej części zębodołowej żuchwy (Pacjentka J.B.).

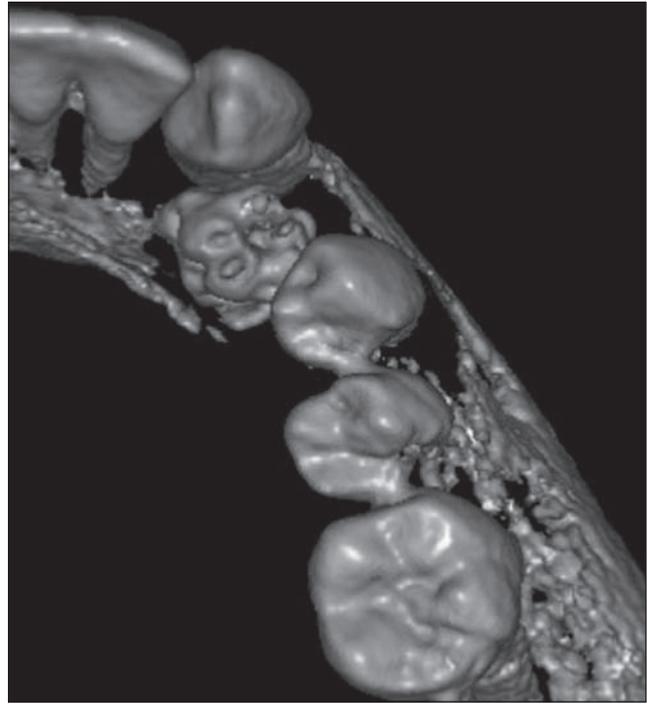


Fig. 3. CBCT scan (patient J.B.).

Ryc. 3. Badanie CBCT (pacjentka J.B.).



Fig. 2. CBCT scan – compound odontoma in the area of teeth 43 and 44.

Ryc. 2. Obraz CBCT – zębiak zestawny w okolicy zębów 43-44.

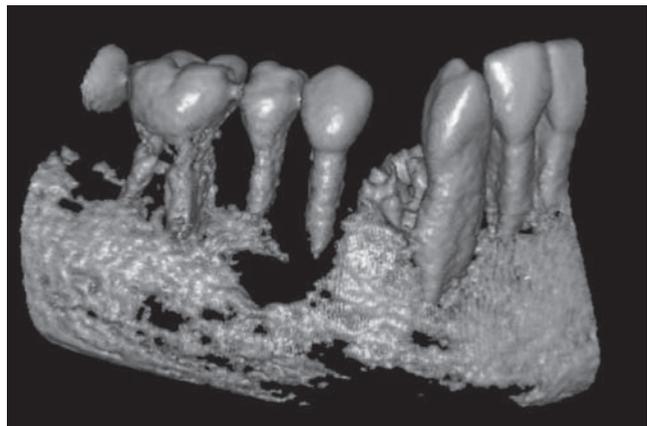


Fig. 4. CBCT scan (patient J.B.).

Ryc. 4. Badanie CBCT (pacjentka J.B.).

The examination revealed a highly mineralised tumour 11mm x 11mm, composed of multiple growths with radiopacity similar to the tooth structures (fig. 2, 3, 4).

The clinical and X-ray examination allowed made it possible to diagnose a compound odontoma in the mandible. After consulting with a surgeon, the decision was taken to remove the lesion during the one -day surgery. The procedure was performed in infiltration and conduction anaesthesia with Lignocainum 2%. A Wassmund cut was used in the area of teeth 43 and 44 to expose the tumour. Once the mucoperiosteal flap had been detached, a fragment of the bone overlaying the odontoma was removed from the lingual side. Then, the lesion composed of 30 odontoids (fig. 5) was excised along with the surrounding capsule, leaving a relatively big cavity (fig. 6). The wound was closed with four Safil 3/0 sutures without filling the cavity with any bone substitute



Fig. 5. Odontoids after enucleation (patient J.B.).

Ryc. 5. Odontoidy (pacjentka J.B.) po wyłuszczeniu.



Fig. 6. Cavity after the procedure (intra-operative photo).  
Ryc. 6. Jama powstała po zabiegu – zdjęcie śródzabiegowe.



Fig. 7. Orthopantomograph (patient J.B.) taken during the therapy with a removable appliance before the CBCT scan and three months after excision of the odontoma.

Ryc. 7. Pantomogram (pacjentka J.B.) wykonany w trakcie terapii aparatem zdejmowanym przed badaniem CBCT i w trzy miesiące po wyłuszczeniu zębiaka.

material. The material was sent for histopathological examination which confirmed the diagnosis of a compound odontoma (fig. 8). After 10 days the wound was declared to heal correctly and the sutures were removed. Follow-up orthopantomographs taken three months later (fig. 7) confirmed that the wound healing process was correct, and further orthodontic treatment with a fixed appliance was planned.

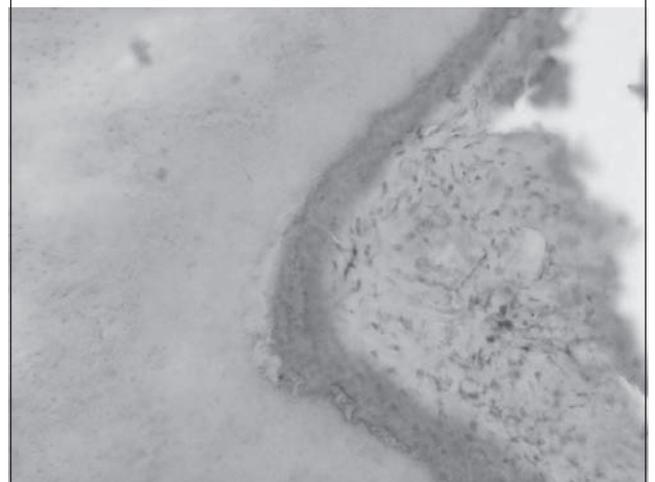


Fig. 8. Histopathological images of fragments from the odontogenic tumour (patient J.B.). Magnification 200x.

Ryc. 8. Obrazy histopatologiczne uzyskane ze skrawków guza zębopochodnego (pacjentka J.B.). Powiększenie 200x.

## DISCUSSION

In our case, the histopathological examination indicated an odontogenic tumour and, combined with the clinical and intraoperative picture, confirmed the diagnosis of a compound odontoma.

Due to the lack of distinct pain symptoms, odontoma-type lesions are usually discovered by chance – in radiographic images taken because of disrupted eruption or dental anomalies in malocclusion [4]. 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 [7]. Their presence may be indicated by bone expansion, slight expanding pain, delayed or arrested eruption of permanent teeth [2] or persistent deciduous teeth [8]. In rare cases, eruption of an odontoma may be accompanied by pain and local swelling [3].

In our case we did not identify persistent deciduous teeth or missing permanent teeth in the mandible, only a gap between the right canine (43) and premolar (44), and distortion of tooth 43. The patient started to feel pain only after the orthodontic therapy with a removable

appliance had been commenced. The pain was caused by the pressure exerted by the plate of the lower part of the appliance, the treatment was painless when the appliance was removed. Probably, the pressure exerted by the appliance activated by the screw might have initiated the eruption of the odontoma, which is an extremely rare case. The lesion was located in the mandible in the area of the first premolar, which is also an unusual site for that type of tumours.

Compound odontomas are usually composed of 4 to 21 odontoids and their size oscillates between 5 mm and 30 mm [9]. In the case described by Jankowska et al, a complex odontoma removed in a teenager was composed of 19 odontoids [10]. Odontomas are usually diagnosed usually in the 2nd decade of life (63%), less often in the 3rd one (24%) [11]. They are usually associated with permanent dentition and appear sporadically in deciduous dentition [12]. In our case the number of odontoids was high, i.e. 30 specimens. Some of them were permanently fused with each other in merged formations or twin odontoids. A similar number of odontoids (37 specimens) was identified by Kubasiewicz-Ros et al [13] in the corresponding location on the opposite site of the mandible and by Niharika et al [14] at a different location (the anterior section of the maxilla).

The treatment consists in complete excision of the tumour. As the lesion is encapsulated, it is possible to dissect the odontoma from the surrounding bone during the surgery. The prognosis is good [1]. Similarly, also in the case of our patient, the odontoma located in the mandible was fully removed, despite the fact that the number of odontoids in the lesion was much higher than in other reported cases of encapsulated growths. In the case of a 25-year-old female patient presented by Kiewlicz et al [4] with a similar location of a complex odontoma, a dozen or so tooth-like growths were discovered after enucleation. The above authors filled the cavity created after the removal of the odontoma. Opinions on that matter vary – some of the authors 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.

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 men more often than women, at the ratio of 7:2. Moreover, in 7 out of 9 cases, the tumour is located in the maxillary alveolar process, especially in the anterior section [15]. The research conducted by Janas in 2000-2007 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. The excised tumours were composed of 7 to 15 odontoids [16].

However, the studies conducted by Bucher demonstrated no diversification by gender as regards the occurrence of odontoma. Similar conclusions were arrived at with respect

to the location, i.e. odontomas occurred equally often in the alveolar area of the mandible and the maxilla. In addition, 62% of odontomas were located in the anterior section of the maxilla [5]. Cases of ectopic odontomas, located in buccal mucosa, were very rare [17].

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. The most common symptom was a missing tooth in the dental arch (55.4% of the cases). As regards the maxillary location, 72.8% of the cases were located in the anterior section, 18.3% in the posterior section and 8.9% in the middle section. In the mandible, 44% of the cases were located in the anterior section, 40.6% in the posterior section and 15% in the middle section [18].

In the presented case presented, we did not determine the aetiological factor, which might have contributed to the compound odontoma composed of such a great number of odontoids. A possible cause might have been a persistent infection in early childhood and the resulting long-term antibiotics therapy, whereas its manifestation and eruption at the age of 11 might have been triggered by the applied orthodontic treatment.

## CONCLUSIONS

1. Odontomas are benign lesions which may be excised during a one-day surgery.
2. The pressure exerted by the plate of a removable appliance is very likely to initiate the eruption of the odontoma.
3. In young patients a slight bone deficiency left after the excision of an odontogenic tumour heals correctly without the need of augmentation with bone substitute material (an this is up to the individual decision of the surgeon).

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**Author's contributions/Wkład Autorów**

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