Giant complex odontoma in the posterior mandible: A case report and literature review

Jong Chan Park, Ji Ho Yang, Sung Youn Jo, Bong Chul Kim, Jun Lee, Wan Lee

Department of Oral and Maxillofacial Surgery, School of Dentistry, College of Dentistry, Wonkwang University, Iksan, Korea

Department of Oral and Maxillofacial Radiology, College of Dentistry, Wonkwang University, Iksan, Korea

ABSTRACT

Odontomas are considered a type of odontogenic hamartoma, and are generally reported not to exceed 3 cm in diameter. Some authors have referred to odontomas with a diameter exceeding 3 cm as giant odontomas. As hamartomas, giant odontomas generally show no signs or symptoms, but if they perforate the mucosa to become exposed in the oral cavity, oral and maxillofacial infections can result. Surgical removal and a histopathological examination may also be required to differentiate them from osteomas, cemento-osseous dysplasia, or mixed odontogenic tumors. This report presents the case of a 28-year-old woman with a giant odontoma in the right mandibular third molar area. Based on a review of the literature published since 2010, only 11 cases of “giant” or “large” odontomas have been reported, most of which were of the complex odontoma type. It was confirmed that they tend to occur in the right posterior mandible. (Imaging Sci Dent 2018; 48: 289-93)

KEY WORDS: Odontogenic Tumors; Odontoma; Hamartoma; Radiography

Case Report

A 28-year-old female patient with schizophrenia presented with pain of a month’s duration in the right mandible. A lesion located in the right aspect of the mandible was observed on a panoramic radiograph taken at a local dental clinic. Following this discovery, the case was referred to Wonkwang University Daejeon Dental Hospital for further management.

In the panoramic view, a calcified mass with a radiolucent rim and heterogeneous radiopacity, together with an impacted third molar, was seen (Fig. 1). On a cone-beam computed tomography (CBCT) scan (Alphard-3030, Asahi Roentgen IND. Co. LTD., Tokyo, Japan; field of view, 10.24 cm; 102 mAs; 80 kV; exposure time, 17 s; and 1...
mm thickness), a radiopaque mass measuring approximately $30 \times 25 \times 20$ mm was found in the right posterior mandibular area with buccal cortical bone expansion and thinning. Additionally, the follicular space of the crown of the impacted third molar was found to be continuous with the radiolucent rim of the radiopaque mass (Fig. 2).

Complex odontoma was diagnosed based on the radiographic findings, because the follicular space of the impacted third molar extended to the boundary of the radiopaque mass. Additionally, it was suspected that the heterogenous radiopacity of the mass was the result of a combination of enamel, dentin, and cementum. Additionally, since the diameter was 3 cm, it was diagnosed as a giant odontoma according to the published literature.

Removal of the giant odontoma and extraction of the left maxillary and left and right mandibular third molars with left iliac bone grafts were done under general anesthesia. During the surgery, the odontoma and impacted right third molar were well separated (Fig. 3). An iliac block bone was fixed in position with 2 mini-plates, and
maxillomandibular fixation (MMF) was performed using a skeletal anchorage system screw and rubber (Fig. 4). The MMF was kept in place for 1 week. After its removal, the specimen was subjected to a histopathological examination, which revealed a mixed pattern of enamel and dentin arranged randomly. This discovery confirmed the diagnosis of complex odontoma (Fig. 5). Removal of the plates was done under general anesthesia 6 months after surgery. The patient was followed up and no major postoperative complications were observed.

Discussion

Odontomas are composed mainly of enamel, dentin, and cementum, and are the most commonly occurring benign odontogenic tumor, accounting for 22% of all such lesions.\(^6,7\) They develop and mature when the permanent teeth are formed, and their development ceases when the permanent teeth are fully matured.\(^3\) The etiology of odontomas is still unclear, but they are presumed to be caused by trauma and infection, genetic factors, or a family history of conditions such as Gardner syndrome and Hermann syndrome.\(^8,9\) Clinically, odontomas are asymptomatic, and are often found incidentally among teenagers. It has been reported that 75.3% of odontomas are detected at routine dental checkups.\(^3\) They occasionally disturb the eruption of deciduous teeth, and roughly 70% of them are associated with impaction, malpositioning, aplasia, malformation, and devitalization of adjacent teeth.\(^10\) Pain and inflammation associated with odontomas have been reported in only 4% of cases.\(^1\)

Odontomas are categorized as compound or complex. Previous studies have shown that compound odontomas are mainly found in the anterior maxillary area, while complex odontomas are found in the posterior mandibular area.\(^11,12\) The sex predilection of this condition is controversial; however, some authors reported that compound odontomas were commoner in males, while complex odontomas were slightly commoner in females.\(^2\) Furthermore, both types of odontomas tend to occur on the right side of both jaws.\(^13\)

Radiographically, complex odontomas are calcified, irregularly-shaped radiopaque masses with narrow radiolucent rims around them and no similarities to the tooth structure. The radiopacity of an odontoma depends on its developmental stage and degree of mineralization. In the first stage, the odontoma is radiolucent due to lack of calcification. In the intermediate stage, slight radiopacity is seen as a result of partial calcification, while in the third stage, radiopacity is increased, with the mass surrounded by a radiolucent rim determined histologically to be connective tissue.\(^3\)

Generally, due to their limited growth, it is rare that the diameter of complex odontomas exceeds 3 cm.\(^4\) In a

![Fig. 3. Clinical photograph showing fragments of a complex odontoma with a left mandibular impacted third molar.](image)

![Fig. 4. A postoperative panoramic image shows that the mass had been removed, the left maxillary and mandibular third molars and the right mandibular third molar had been extracted, and the left iliac block bone had been fixed with mini-plates. Maxillomandibular fixation was performed using a skeletal anchorage system screw and rubber.](image)
study by Miki et al., only 4.3% of odontomas were found to be larger than 3 cm. The terms “giant odontoma” and “large odontoma” were used to search Google Scholar and PubMed for reports published since 2010. This search retrieved 11 cases that provided data on the sizes of the odontomas (Table 1). A further review showed that only 8 of the cases of reported giant odontomas with diameters exceeding 3 cm. In these studies, giant odontomas tended to be located on the right (9 of 11), and in the mandibular (9 of 11), and posterior (8 of 11) areas. Furthermore, most giant odontomas were classified as complex odontomas (7 of 11), and they occurred mainly in patients under 30 years of age (9 of 11).

In our case, the complex odontoma measured $3 \times 2.5 \times 2$ cm, which is similar to the size of the odontoma described by Vengal et al. A giant odontoma measuring up to 6 cm has been reported, with the largest odontoma weighing 0.3 kg. The abnormal size of the lesion resulted in expansion of the jaws and facial asymmetry. Giant odontomas rarely perforate the oral mucosa, and when they do, they are referred to as sequestrating or erupting odontomas. When this happens, pain, inflammation, and infection of the adjacent oral structures, such as bone, adjacent teeth, and gingiva occur. Furthermore, in our case, the radiolucent area surrounding the crown of the impacted third molar, which was suspected to be the dental follicle, was continuous with the radiolucent rim of the giant odontoma. Through this radiographic evaluation, it was determined that the giant odontoma developed from the malformed dental epithelium of the impacted third molar. Additionally, the third molar was severely impacted, judging from the malpositioning of the adjacent tooth, and this impaction resulted from the presence of the giant odontoma.

Based on the radiolucent rim around the radiopaque mass on the radiograph, the differential diagnoses considered included osteoma and cemento-osseous dysplasia (COD). Other mixed odontogenic tumors such as ameloblastic fibro-odontoma or odontoameloblastoma, which grow aggressively, were also considered. Radiographically, osteomas are well-demarcated, ovoid, radiopaque lesions that can be distinguished from complex odontomas by the lack of a radiolucent rim around the lesion. It is easy for CODs in the end stage to be confused with complex odontomas because they appear as dense radiopaque masses with radiolucent rims on radiographs. However, CODs occur in persons over 30 years of age and are related to the root, whereas complex odontomas occur mainly in adolescents and are located in the alveolar bone of the jaw. Ameloblastic fibro-odontomas present as well-demarcated radiolucencies containing radiopaque material and usually occur in patients younger than 20 years of age. Odontoameloblastomas, in con-

---

### Table 1. Published cases of giant odontomas since 2010.

| Authors          | Size          | Position        | Age  | Sex | Type     |
|------------------|---------------|-----------------|------|-----|----------|
| Bagewadi et al.  | $4 \times 3.5$ cm | Rt. Mn. Posterior | 22   | M   | Complex  |
| Spini et al.     | 6 cm          | Mn. Anterior    | 9    | M   | Complex  |
| Perumal et al.   | $5.5 \times 4 \times 2.5$ cm | Rt. Mn. Posterior | 24   | F   | Complex  |
| Lee et al.       | $2.7 \times 1.0 \times 1.8$ cm | Rt. Mx. Anterior | 41   | F   | Compound |
|                  | $2.3 \times 2.0 \times 1.9$ cm | Rt. Mn. Posterior | 16   | F   | Compound |
| Chrcanovic et al.| $4 \times 3 \times 2.75$ cm | Rt. Mn. Posterior | 21   | F   | Complex  |
| Utumi et al.     | 4 cm          | Rt. Mx. Anterior | 10   | F   | Complex  |
| Akerzoul et al.  | $6 \times 6$ cm | Lt. Mn. Posterior | 35   | F   | Compound |
| Lehman et al.    | $8 \times 4$ cm | Rt. Mn. Posterior | 7    | F   | Compound |
| Biocie et al.    | $5 \times 3$ cm | Rt. Mn. Posterior | 10   | F   | Complex  |
| Reddy et al.     | $2.9 \times 1.6$ cm | Rt. Mn. Posterior | 13   | M   | Complex  |

Rt.: right, Lt.: left, Mx.: maxilla, Mn.: mandible
trans, are characterized by multilocular radiolucent lesions with radiopaque areas resembling dental tissue, and also predominantly affect patients younger than 20 years of age.\textsuperscript{21} The main difference between complex odontomas and mixed odontogenic tumors is their growth potential, as exemplified by the fact that even an odontoameloblastoma can cause root resorption of the adjacent teeth.

Although an odontoma is a hamartoma, its recommended treatment involves conservative surgical removal and histopathological examination of the mass to differentiate it from other lesions.\textsuperscript{22} Very few cases of recurrence after removal have been recorded, and the prognosis is excellent.\textsuperscript{23}

This report has provided a better understanding of the clinical and radiological features of complex odontomas. 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. Its accurate diagnosis is important, and conservative surgery is the most appropriate treatment.

References

1. Bagewadi SB, Kukreja R, Suma GN, Yadav B, Sharma H. Unusually large erupted complex odontoma: a rare case report. Imaging Sci Dent 2015; 45: 49-54.
2. Hidalgo-Sánchez O, Leco-Berrocal MI, Martínez-González JM. Metaanalysis of the epidemiology and clinical manifestations of odontomas. Med Oral Patol Oral Cir Bucal 2008; 13: E730-4.
3. An SY, An CH, Choi KS. Odontoma: a retrospective study of 73 cases. Imaging Sci Dent 2012; 42: 77-81.
4. Spini PH, Spini TH, Servato JP, Faria PR, Cardoso SV, Loyola AM. Giant complex odontoma of the anterior mandible: report of case with long follow up. Braz Dent J 2012; 23: 597-600.
5. Perumal CJ, Mohamed A, Singh A, Noffke CE. Sequestrating giant complex odontoma: a case report and review of the literature. J Oral Maxillofac Surg 2013; 12: 480-4.
6. Kulkarni VK, Vanka A, Shashikiran ND. Compound odontoma associated with an unerupted rotated and dilacerated maxillary central incisor. Contemp Clin Dent 2011; 2: 218-21.
7. Lee J, Lee EY, Park EJ, Kim ES. An alternative treatment option for a bony defect from large odontoma using recycled demineralization at chairside. J Korean Assoc Oral Maxillofac Surg 2015; 41: 109-15.
8. Chrzanovic BR, Jaeger F, Freire-Maia B. Two-stage surgical removal of large complex odontoma. Oral Maxillofac Surg 2010; 14: 247-52.
9. Utumi ER, Cremonini CC, Pedron IG, Zambon CE, Cavalcanti MG, Cecchetti MM. Maxillary reconstruction with particulate bone graft and titanium mesh: a treatment option for large complex odontoma of the maxilla. J Dent Child (Chic) 2011; 78: 124-8.
10. Bodin I, Julin P, Thomsson M. Odontomas and their pathological sequels. Dentomaxillofac Radiol 1983; 12: 109-14.
11. Amado Cuesta S, Gargallo Albíol J, Berini Aytés L, Gay Escoda C. Review of 61 cases of odontoma. Presentation of an erupted complex odontoma. Med Oral 2003; 8: 366-73.
12. Akerzoul N, Chbicheb S, El Wady W. Giant complex odontoma of mandible: a spectacular case report. Open Dent J 2017; 11: 413-9.
13. Baldawa RS, Khante KC, Kalburge JV, Kasat VO. Orthodontic management of an impacted maxillary incisor due to odontoma. Contemp Clin Dent 2011; 2: 37-40.
14. Miki Y, Oda Y, Iwaya N, Hirota M, Yamada N, Aisaki K, et al. Clinicopathological studies of odontoma in 47 patients. J Oral Sci 1999; 41: 173-6.
15. Vengal M, Arora H, Ghosh S, Pai KM. Large erupting complex odontoma: a case report. J Can Dent Assoc 2007; 73: 169-73.
16. Ragalli CC, Ferreria JL, Blasco F. Large erupting complex odontoma. Int J Oral Maxillofac Surg 2000; 29: 373-4.
17. Lehman H, Lustmann J, Regev E. Removal of an extensive mandibular odontoma using an intraoral approach. Quintessence Int 2013; 44: 425-8.
18. de França TR, Gueiros LA, de Castro JF, Catunda I, Leão JC, da Cruz Perez DE. Solitary peripheral osteomas of the jaws. Imaging Sci Dent 2012; 42: 99-103.
19. Biocic J, Macan D, Brjadic D, Manojlovic S, Butorac-Rakvin L, Hat J. Large erupting complex odontoma in a dentigerous cyst removed by a piecemeal resection. Pediatr Dent 2010; 32: 255-9.
20. Mortazavi H, Baharvand M. Jaw lesions associated with impacted tooth: a radiographic diagnostic guide. Imaging Sci Dent 2016; 46: 147-57.
21. Dive A, Khandekar S, Bodhade A, Dohbley A. Odontoameloblastoma. J Oral Maxillofac Pathol 2011; 15: 60-4.
22. Reddy GS, Reddy GV, Sidhartha B, Sriharsha K, Koshy J, Sultana R. Large complex odontoma of mandible in a young boy: a rare and unusual case report. Case Rep Dent 2014; 2014: 854986.
23. Tomizawa M, Otsuka Y, Noda T. Clinical observations of odontomas in Japanese children: 39 cases including one recurrent case. Int J Paediatr Dent 2005; 15: 37-43.
