

Management of Large Odontoma in Mandible: A Rare Case Report

Yuthakran Aschaitrakool¹, Tin Panyawaraphon¹, Chonnatee Chinkrua¹, Nutchapon Chamusri²,
Yuniardini Septorini Wimardhani³

¹Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Chiang Mai University, Thailand

²Department of Oral Biology and Oral Diagnostic Sciences, Faculty of Dentistry, Chiang Mai University, Thailand

³Department of Oral Medicine, Faculty of Dentistry, Universitas Indonesia, Indonesia

Received: April 10, 2021 • Revised: May 16, 2021 • Accepted: May 21, 2021

Corresponding Author: **Yuthakran Aschaitrakool** Lecturer, Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Chiang Mai University, Chiang Mai 50200, Thailand. (E-mail: yuthakran.a@cmu.ac.th)

Abstract

Odontomas are considered to be developmental anomalies (hamartomas), rather than true neoplasm, composing of both epithelial cells and mesenchymal dental hard tissues. The prevalence of odontoma at 20.1 % is regarded as the second most common tumor occurring in the oral cavity. Based on pathological conditions, odontomas are classified into two types, compound odontomas and complex odontomas. This study investigates the case of a 19-year-old Thai female who was referred to the department of oral and maxillofacial surgery at Chiang Mai University and presented with a swollen right cheek that developed gradually. Without any previous symptoms, the tumor was accidentally detected from a radiograph during an orthodontic treatment in a private clinic. The patient's past medical history showed no underlying disease or drug allergy. The clinical examination revealed facial asymmetry with swelling present on the right buccal area, normal skin texture, no tenderness, and no abnormal sensation. An intraoral investigation revealed a bony-hard swelling of the right mandible from distal of the mandibular right second molar to the ramus of mandible; however, the mandibular right third molar was not found during the clinical examination. The panoramic radiograph exhibited a unilocular nonhomologous radiopaque lesion with a well-defined margin and an expanded border to the right mandible. The inferior alveolar canal and the mandibular right third molar were displaced inferiorly to the inferior border of the mandible. An incisional biopsy was performed under local anesthesia and the diagnosis showed it was complex odontoma. The odontoma was enucleated by extraoral approach. After the lesion had been removed, internal fixation was done using a reconstruction plate. The patient had no pain or postoperative paresthesia during a 12-month follow-up period. As such, the removal of the reconstruction plate was carried out 18 months after the surgery. In conclusion, this case is a rare incidence of large odontomas at the right angle of the mandible. Despite its large size, conservative treatment with a reconstruction plate to prevent a pathologic fracture yielded satisfactory results, which is consistent with other studies in literature.

Keywords: large odontoma, odontoma, reconstruction plate

Introduction

Odontomas are classified as developmental anomalies (hamartomas), rather than true neoplasm, with stem cells derived from ectomesenchyme, which is associated with the creation of various tooth components such as enamel, dentine, pulp, and cementum. Odontomas are usually found accidentally in a radiograph from oral health examination or detected from a gradual swelling of the jaw bone in the diseased area, and are typically presented without any symptoms.^(1,2) However, swelling and pain are the common symptoms that may occur in association with other odontogenic tumors, most notably calcifying odontogenic cyst and dentigerous cysts.⁽³⁾

The prevalence of odontoma was found to be 20.1%, and is regarded as the second most common tumor occurring in the jaw bone.⁽¹⁾ From the literature review, odontomas are generally small, with an average size of approximately 1.9-3.4 cm.^(4,5) Only a few of them were found to be larger than 6 cm in diameter and even fewer cases of large-size odontoma occurring in the jaw have been reported.^(1,6-8)

The conservative treatment approach is advisable for odontoma depending on the location and size of tumor present in each patient.⁽¹⁾

The objective of this study is to report a case with large odontoma in the mandible who received treatment at the Faculty of Dentistry, Chiang Mai University.

Case report

A 19-year-old Thai woman came to see a dentist at Oral and Maxillofacial Surgery Clinic, Faculty of Dentistry, Chiang Mai University with a swollen right cheek as the main symptom. According to the patient, her right cheek had been gradually swelling without any pain for a few years and was accidentally detected from a radiograph during orthodontic treatment in a private clinic. Past medical history revealed no underlying disease or drug allergy. From the external oral examination, the face was asymmetric with a mild swell on the right cheek. The skin looked normal, and no pain, tenderness or numbness were felt or reported. Additionally, no enlarged lymph nodes were detected. Besides, the patient could open her mouth at normal wideness (Figure 1).

The oral examination showed an enlargement of bones in the alveolar part from the distal aspect of the mandibular right second molar to the anterior of the ramus bone in the mandible, which was a bony hard and swelling but without pain. After the sensibility test, the mandibular right second molar had positive signs and there were prints of tooth bites from the maxillary right third molar at the alveolar mucosa distal to the mandibular right second molar. In the oral cavity, while none of the teeth were decayed, the mandibular right third molar was not found (Figure 2).



Figure 1: shows facial asymmetry with swollen right cheek, normal skin, and no lymphadenopathy.



Figure 2: shows bony hard swelling with buccal and lingual bone expansion on the right mandible. The mandibular right third molar is clinically absent.

From a panoramic radiograph and Postero-Anterior skull radiographs, a large radiopaque mass was presented in the right mandible with an approximate size of 4x3 cm. The mass expanded in the anterior-posterior aspect, from the distal aspect of the mandibular right second molar to the ramus of the mandible, as well as in the vertical aspect, from the alveolar part to the inferior border of the mandible. Furthermore, the radiograph showed bucco-lingual expansion of the bone together with an embedded mandibular right third molar, which was displaced to the inferior border of mandible by the lesion. From the periapical radiograph, there was no root resorption of the mandibular right second molar (Figure 3).

Cone-beam computed tomography was further examined to display the varies cross-section of the mandible, and the heterogeneous opacity lesion with clear boundaries was presented. A bucco-lingual expansion of the jawbone was also found to have destroyed the cortical bone and perforated some part of the lingual cortex of the mandible. The lesions displaced the inferior alveolar nerve

and the embedded of mandibular right third molar. There was no root resorption of the mandibular right second molar (Figure 4).

The original diagnosis of this condition was odontoma, ossifying fibroma calcifying odontogenic cyst and calcifying epithelial odontogenic tumor. The final diagnosis was obtained after a biopsy. Oral and maxillofacial surgeons performed an incisional biopsy at the lesion. The histopathological features of the hard tissue cut from the lesion appeared like many fragments of teeth (Figure 5).

The patient underwent extraoral surgery after being given general anesthesia at the submandibular area to remove odontoma and the embedded mandibular right second molar before fixing with a reconstruction plate and screws (Figure 6). Subsequently, the histopathological diagnosis of complex odontoma was made. After a 12-month follow-up post the surgery, the patient's wound had completely healed with no pain or numbness, and the mandibular right second molar remained alive, as the patient continued receiving orthodontic treatment. Eigh-

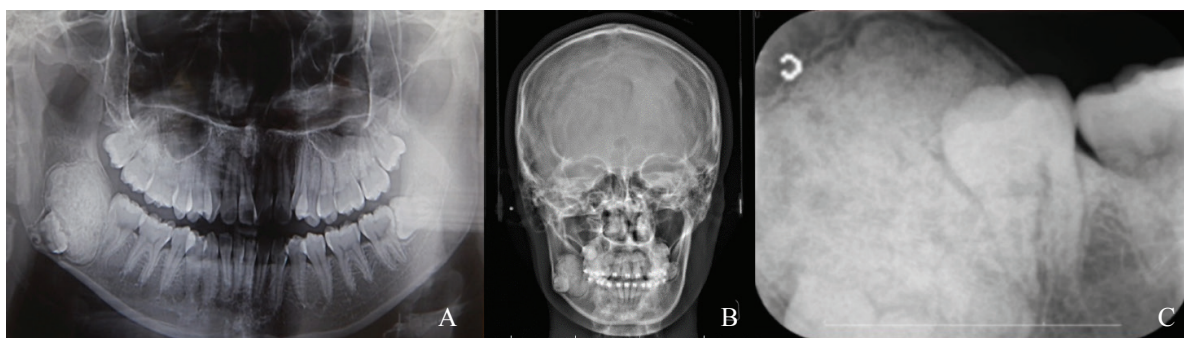


Figure 3: shows (A) panoramic, (B) PA skull, (C) periapical radiograph with a well-defined amorphous radiopaque mass at the right mandible. The mandibular right third molar and inferior alveolar canal are displaced. The mandibular right second molar has intact lamina dura and no root resorption.

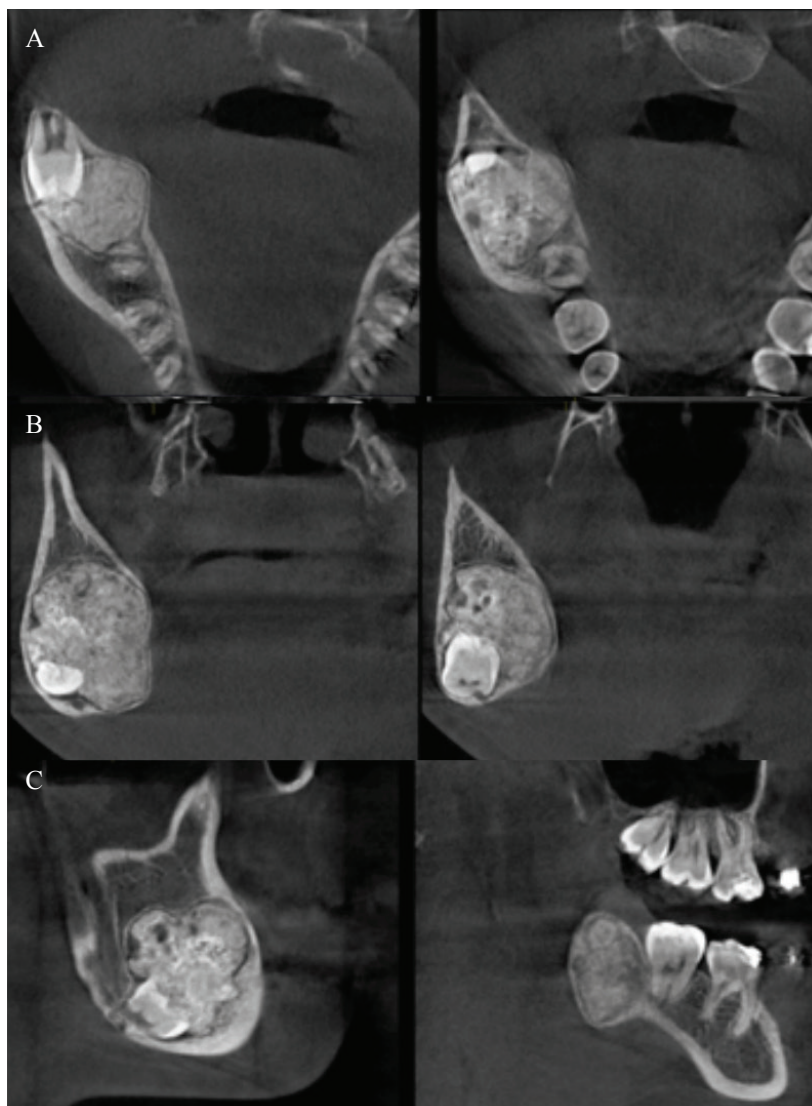


Figure 4: shows (A) axial, (B) coronal, and (C) sagittal CBCT images displaying a well-defined amorphous radiopaque mass with bucco-lingual expansion. The lesion displaces the inferior alveolar canal and the mandibular right third molar. Root resorption of mandibular right second molar is not observed.

teen months following the surgery, the patient underwent another surgery under general anesthesia to remove the reconstruction plate and screws.

Discussions

According to the World Health Organization, odontomas are presently classified into two types based on pathological conditions: compound odontomas and complex odontomas.⁽⁹⁾ While both are hard tissues and similar to the structure of teeth including enamel, dentin, enamel root, and pulpal tissues surrounded by connective tissues, the hard tissues of compound odontomas are arranged

orderly similar to natural teeth as opposed to those of complex odontomas which look dissimilar to natural teeth and exhibit disorderly arrangement.^(1,2,10)

Radiographic characteristics generally present as radiopaque lesion surrounded by a halo. Odontoma radiographs can be divided into 3 stages according to the progress of lesion development:

1. Stage 1: Opacity radioactivity indicating undeveloped hard tissues
2. Stage 2: Radiopaque occurring from partial calcification
3. Stage 3: Radiopaque surrounded by a thin halo, which is mostly found

The radiographic pictures of compound odontoma normally display a radiopaque shadow in tooth-like structure whereas the pictures of complex odontoma usually reveal irregularly shaped and unspecified radiopaque mass.⁽¹¹⁾

Odontomas are usually found between the ages of 1-30 years, with equal incidence rates for males and females. These mostly occur in the mandible rather than the maxilla, and in the posterior teeth more than the anterior teeth. Complex odontomas generally present in the posteri-

or teeth; however, compound odontomas are typically found among young patients and in the anterior region of the jaw.⁽¹²⁾ While the pathogenesis is unknown, possible causes of odontomas include traumatic injuries to primary teeth, inheritance, and genetic abnormalities. In addition, certain anomalies such as Gardner syndrome, Hermann syndrome, familial colonic adenomatosis, and basal cell nevus are also associated with the incidence of odontomas.⁽¹³⁾

While odontomas are usually small, large-sized odontomas have also been found. The larger odontomas may cause an expansion of the jawbone resulting in an asymmetrical face. In the maxilla, large odontomas can

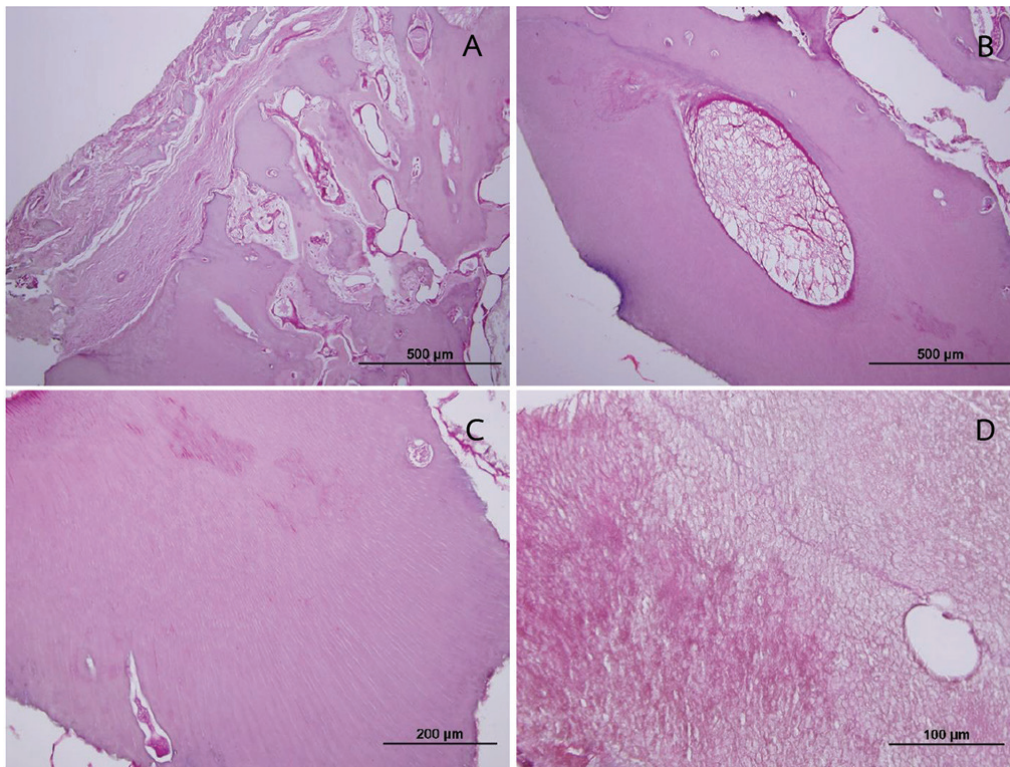


Figure 5: shows histopathological images. (A) Fibrous connective tissue surrounding the lesion (H&E, x100) (B) Dentin-pulp complex (H&E, x100) (C) Calcified bodies resembling dentin (H&E, x200) (D) Enamel matrix (H&E, x400)

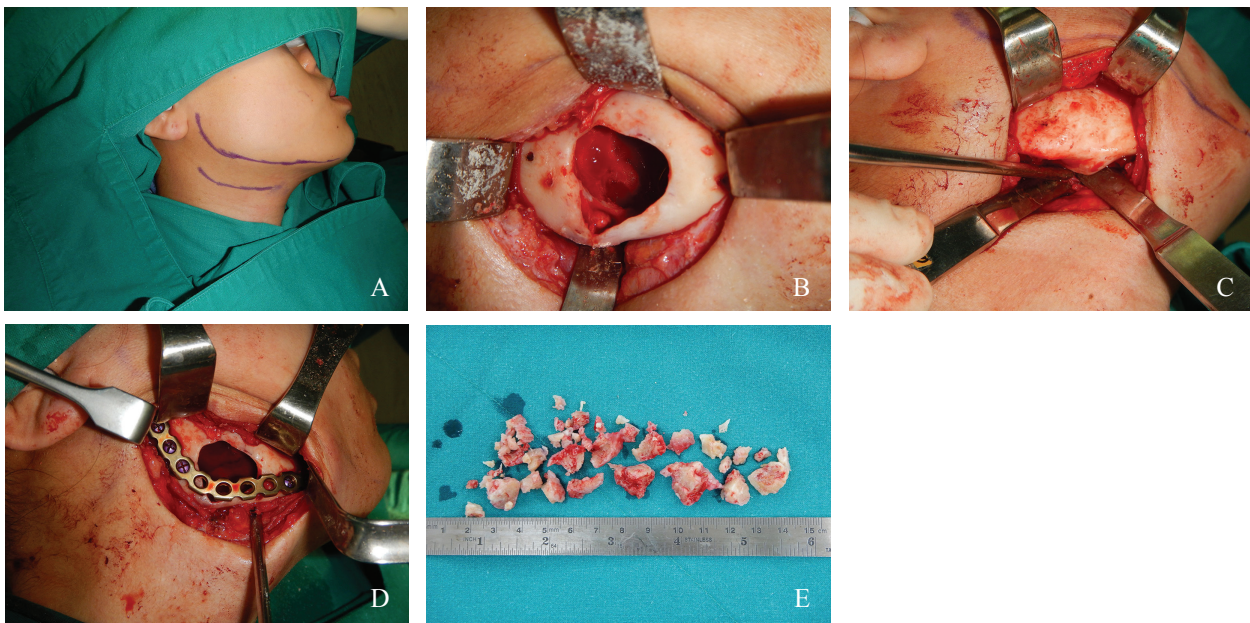


Figure 6: shows perioperative images. (A) Submandibular approach as an extraoral incision 2 inches below the mandible. (B) Soft tissue dissection through bony part of the mandible. (C) Buccal corticotomy at the lesion for enucleation and surgical removal of the mandibular right second molar. (D) Fixation with reconstruction plate and screws. (E) Gross specimens consisting of numerous pieces of hard tissue from the entire lesion.

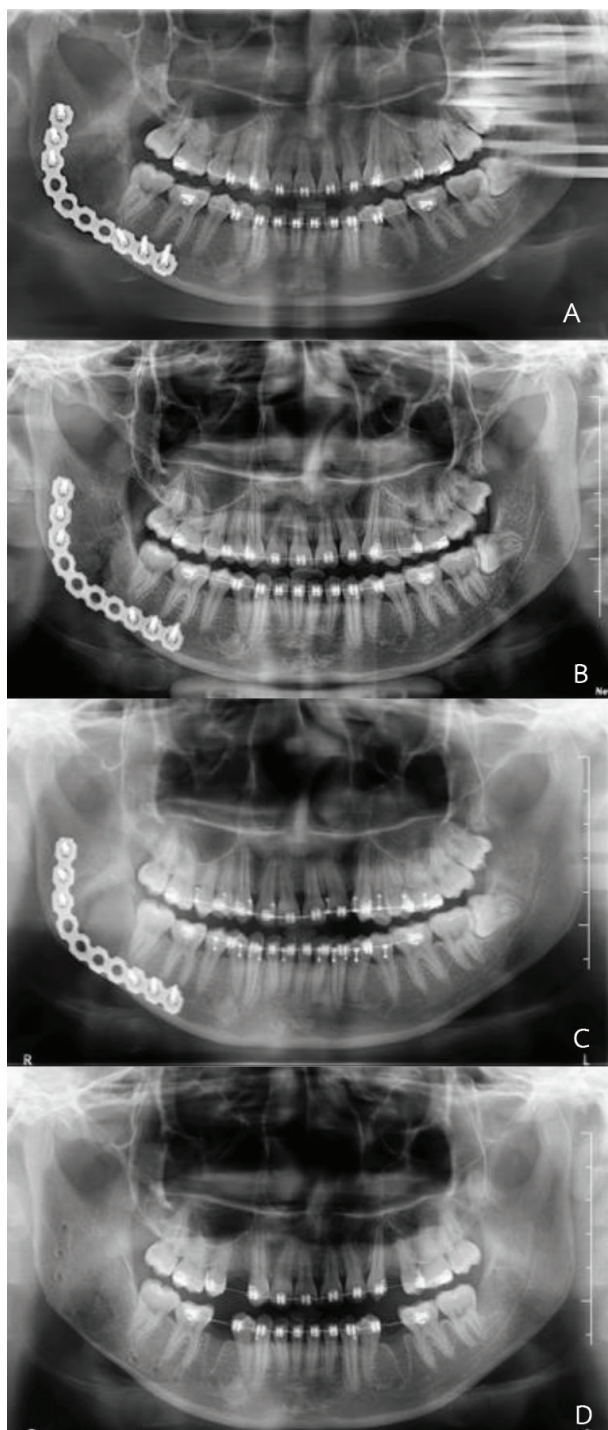


Figure 7: shows postoperative panoramic radiographs. (A) 3 months after surgery (B) 6 months after surgery (C) 12 months after surgery (D) after reconstruction plate removal

occur in the nasal cavity but rarely cause inflammation and infection in the ventricles if the lesions do not erupt in the mouth.^(14,15) The large mass might also impact the adjacent teeth, usually with no incidence of root resorption.^(6-8,16,17) In addition, odontomas are often associated with embedded teeth which can be displaced away from the original position by the large mass.^(6,8) Some studies suggest that surgical removal of the mass could allow the embedded teeth to erupt.⁽¹⁸⁾ Therefore, detecting the lesion at a very young age is best for the patient. With respect to this case, the patient demonstrated general characteristics of odontomas that exhibited no symptoms and were accidentally found at the annual oral examination.^(15,17) Although the detected odontoma was large, a mild characteristics of malignant tumor-like lesion with no pain and numbness was present. Several studies indicate that the pain symptom among patients with large odontoma is usually due to the infection of the lesion emerging through the gum. These patients tend to see the dentist with sudden acute swelling and pain, and lesions can be clearly seen with infection during the oral examination.^(6,8,19,20)

Differential diagnosis is an important part of treatment planning. Most patients are asymptomatic, and the lesions are accidentally detected by routine radiograph. Ossifying fibroma, calcifying odontogenic cyst, and calcifying epithelial odontogenic tumors are considered due to radiopaque occurring from partial calcification or radiopaque surrounded by a thin halo. However, odontoma are usually small and painless, and with higher incidence of occurring in the jawbone.

General treatment for odontomas is administered conservatively by removing the mass under minor surgery since the lesions are typically small, slow-growing, and non recurrent.^(1,2) Various treatment modalities have been described for the treatment of odontoma where either an extraoral approach or intraoral approach have been reported as surgical techniques for the removal of odontoma. The size, location, extension of the lesion, histological aspects of odontoma, and age of the patient are important factors which have to be considered while planning the surgery.⁽³⁾ In this patient, however, the detected lesion showed a large bone expansion of the inferior border of the mandible, which results in extensive bone loss after the surgery. In consequence, the extraoral approach under general anesthesia was performed before fixing the defective bone with

a reconstruction plate and screws to prevent the pathologic fracture of the mandible. Accordingly, Akbulut, in 2018, treated a case of large complex odontoma of the right angle of the mandible by using both intraoral and extraoral surgical methods and reconstruction was done with titanium reconstruction plates.⁽²¹⁾

As this patient was undergoing orthodontic treatment, there was concern for bones surrounding adjacent tooth roots beside the lesion area. Many studies found that large odontomas show a good prognosis in both the upper and lower jaws.^(7,8,14,15,22) One case study suggested processing hard tissues of odontomas before inserting them back into the bone cavity as an optional of bone substitutes material. Despite its favorable result, further comprehensive studies are required.⁽¹⁶⁾ During 12-month follow-up with panoramic radiography after the surgery, new bones have been formed in the lesion area. The reconstruction plate and screws were then removed and orthodontic treatment was continued until its completion.

Regarding the diagnosis, histopathology is essential in order to differentiate large-sized odontomas from other more severe lesions that require more aggressive treatment such as calcifying epithelial odontogenic tumor and ossifying fibroma.^(1,2)

Conclusions

This study reports a patient with large complex odontomas in the mandible who received treatment at the Faculty of Dentistry, Chiang Mai University. Large-sized odontomas are not frequently found, and need to be removed from extraoral site by a general anesthesia surgery before securing the bones with a reconstruction plate and screws. The type of surgical approach may be considered as either conservative or radical including reconstruction procedure according to their size and remaining bone quantity. Pathologic fracture of the mandible should be a concern due to management of expanded lesion with very thin cortical plate remaining. The clinical features, radiographs, and histopathology of this lesion conformed with other studies reviewed from previous literature.

References

1. Neville BW, Damm DD, Allen CM, Chi AC. *Oral and maxillofacial pathology*. 4th ed. St. Louis, MO: Elsevier; 2016: 674-675.
2. Nattapong V. *Oral pathology: surgical differential diagnosis*. 1st ed. Bangkok: Department Of pathology oral and maxillofacial surgery Faculty of dentistry Mahidol University; 2014: 262-267.
3. Saravanan R, Sathyasree V, Manikandhan R, Deepshika S, Muthu K. Sequential removal of a large odontoma in the angle of the mandible. *Ann Maxillofac Surg* 2019; 9(2): 429-433.
4. de Medeiros WKD, da Silva LP, Santos PPA, Pinto LP, de Souza LB. Clinicopathological analysis of odontogenic tumors over 22 years period: Experience of a single center in northeastern Brazil. *Med Oral Patol Oral Cir Bucal* 2018; 23(6): 664-671.
5. Mehngi R, Rajendra K, Bhagwat P, Hegde SS, Sah D, Rathod VS. Clinical and histopathological analysis of odontogenic tumors in institution-a 10 years retrospective study. *J Contemp Dent Pract* 2018; 19(10): 1288-1292.
6. Vengal M, Arora H, Ghosh S, Pai KM. Large erupting complex odontoma: a case report. *J Can Dent Assoc* 2007; 73(2): 169-173.
7. 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(5): 597-600.
8. Perumal CJ, Mohamed A, Singh A, Noffke CE. Sequestering giant complex odontoma: a case report and review of the literature. *J Maxillofac Oral Surg* 2013; 12(4): 480-484.
9. Soluk-Tekkeşin M, Wright JM. The World Health Organization Classification of Odontogenic Lesions: A Summary of the Changes of the 2017 (4th) Edition. *Turk Patoloji Derg* 2013; 34(1).
10. Slootweg PJ. Lesions of the jaws. *Histopathology* 2009; 54(4): 401-418.
11. Ongole R, BN P. *Textbook of oral medicine, oral diagnosis and oral radiology*. Haryana: Elsevier; 2013.
12. Soluk-Tekkeşin M, Pehlivan S, Olgac V, Aksakalli N, Alatlı C. Clinical and histopathological investigation of odontomas: review of the literature and presentation of 160 cases. *J Oral Maxillofac Surg* 2012; 70(6): 1358-1361.
13. Katz RW. An analysis of compound and complex odontomas. *ASDC J Dent Child* 1989; 56(6): 445-449.
14. Murphy C, O'Connell JE, Cotter E, Kearns G. Management of large erupting complex odontoma in maxilla. *Case Rep Pediatr* 2014; 2014: 963962. doi:10.1155/2014/963962
15. Subbalekha K, Tasanapanont J, Chaisuparat R. Giant Complex Odontoma: A Case Report. *The Bangkok Medical Journal* 2015; 9(1): 50-56.

16. 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(2): 109-115.
17. Akerzoul N, Chbicheb S, El Wady W. Giant Complex Odontoma of Mandible: A Spectacular Case Report. *Open Dent J* 2017; 11: 413-419.
18. Isola G, Cicciu M, Fiorillo L, Matarese G. Association between odontoma and impacted teeth. *J Craniofac Surg* 2017; 28(3): 755-758.
19. Bagewadi SB, Kukreja R, Suma GN, Yadav B, Sharma H. Unusually large erupted complex odontoma: a rare case report. *Imaging Sci Dent* 2015; 45(1): 49-54.
20. D'Cruz AM, Hegde S, Shetty UA. Large Complex Odontoma: a report of a rare entity. *Sultan Qaboos Univ Med J* 2013; 13(2): 342-345.
21. Akbulut N. Huge Complex odontomas in two cases with unique history and treatment protocols. *Clin Surg* 2018; 3: 1888.
22. Park JC, Yang JH, Jo SY, Kim BC, Lee J, Lee W. Giant complex odontoma in the posterior mandible: a case report and literature review. *Imaging Sci Dent* 2018; 48(4): 289-293.