Case Report - Cyst and Tumours

Bilateral Massive Complex Composite Odontoma of the Mandible: A Rare Case Report

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Abstract

Rationale: Odontomas result from abnormal growth of differentiated epithelial and mesenchymal cells, which form ameloblasts and odontoblasts along with pulp tissue at times. Patient Concerns: A patient reported with a chief complaint of hard swelling of the lower jaw causing difficulty in mouth opening and facial deformity. Diagnosis: A provisional diagnosis of complex odontome based on clinicoradiological findings was made, which was later confirmed as complex composite odontome histologically. Treatment: Bilateral hard tissue mass was surgically removed from the mandible with utmost precaution to prevent jaw deformity. On fine separation, the specimen yielded nearly 82 small teeth-like structures. Outcomes: Histopathologically, multiple small teeth-like elements with haphazardly arranged central fibrofatty pulpal stroma surrounded by well-formed dentinal tubules and empty areas representing decalcified enamel matrix surrounded by fibrovascular stroma at the periphery were seen. Take-away Lessons: Early diagnosis and treatment ensures minimal surrounding deformities, better prognosis, and less chances of recurrence.

Keywords: Complex, composite, compound, hamartoma, odontoma

Introduction

Odontomas are hamartomas, resulting from abnormal growth of absolutely differentiated epithelial and mesenchymal cells, which form ameloblasts and odontoblasts.[1] Broca in 1866 coined the term odontoma and defined it as a tumour of overgrowth of complete dental tissues such as enamel and dentin with considerable amounts of cementum and pulp tissue also at times.[2] Odontomas are nonaggressive, benign developmental anomalies, which are passive in growth[3] and seldom exceed the size of a tooth, but when large, it may cause expansion of the cortical bone.[4] They are considered perfidious neoplasms because even being deficient in structural arrangement, the epithelial and mesenchymal cells of an odontoma may appear normal.[5] They may be histologically classified as compound if the lesion has an apparent anatomic similarity to elementary teeth arranged in an orderly pattern, featuring multiple small teeth-like elements called odontoids or denticles or as complex when the calcified dental tissues appear simply as an irregular mass composed mainly of mature tubular dentin in a disorderly pattern.[6] Complex odontomas are rarer than the compound variety in the ratio of 1:2.[2]

Although of an unknown etiology, local trauma, infection, and genetic factors affecting extraneous buds of odontogenic epithelial cells may be attributed to its etiology.[7] Odontomas located in the anterior region of the maxilla are mostly compound while the majority of odontomas located in the posterior areas, especially in the mandible, are complex odontomas.[8] Complete surgical excision is the main mode of management with a favorable prognosis complying scanty chances of recurrence.[9] In this article, we report a rare bilateral massive complex composite odontoma of the mandible.

Case Report

A 17-year-old male patient was referred to the craniomaxillofacial unit with a chief complaint of large hard swelling of bilateral

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lower jaw, leading to difficulty in mouth opening and consistent mild pain for 5 years. His medical and family history was insignificant. There was no history of trauma, infection or swelling of the mandible and maxillofacial region before the initiation of his current facial deformity. On extraoral examination, a bony hard swelling of size 3.5 cm × 4.8 cm on the right side and 3.3 × 5.1 cm on the left side approximately was evaluated, which was nonmobile, nonpulsatile, nontender, and nonulcerative in nature [Figure 1]. The patient’s mouth opening was near normal, but difficulty in mouth opening was assessed due to bulky bilateral bony swelling. On intraoral examination, the occlusion was normal and a bilateral hard swelling was palpable linguually extending up to floor of the mouth with elevation of lateral border of tongue, assessed as extension of the extraoral bony swelling. The orthopantomogram radiographic picture appeared as a bag of multiple irregular masses of calcified material surrounded by a thin radiolucent area with smooth periphery of size 4 cm × 4.5 cm approximately, involving the posterior base and angle of the mandible bilaterally, along with many other well-formed impacted permanent teeth [Figure 2]. The coronal and sagittal sections of contrast-enhanced computed tomography revealed bilateral multiple large nonspecific, disorganised, irregular radio-opaque masses with varying densities, unable to be recognised as dental tissue [Figure 3]. Extended Risdon’s extraoral surgical approach was utilised and bilateral hard tissue mass was explored [Figure 4]. The hard tissue masses were gently chiseled from buccal and lingual sites bilaterally, and on the left side, while removing the hard tissue mass, an iatrogenic mandibular body fracture occurred, which was reduced and fixed with a titanium miniplate [Figure 5]. The remaining well-formed impacted permanent teeth were left considering their delayed eruption. Specimen collected was large chunks of hard tissue masses with tooth-like structures in it, which on fine separation yielded nearly 82 small teeth-like structures [Figure 6]. The histopathological section revealed multiple small teeth-like structures with haphazardly arranged central fibrofatty pulpal stroma surrounded by well-formed dentin showing dentinal tubules and empty areas representing decalcified enamel matrix surrounded by fibrovascular stroma at the periphery, and a diagnosis of complex composite odontoma was made [Figure 7]. A 3-month follow-up showed uneventfully healed surgical site without any jaw deformity and radiologically intact shape of the mandible with healed fracture site on the left side body of the mandible [Figure 8].

**Discussion**

The incidence of compound odontome ranges from 9% to 37% and most of odontomas in the anterior region of the jaws are compound composite (61%), whereas those in the posterior segment are complex composite in nature (34%). Most both types of odontomas often occur more on the right side (compound 62% and complex 68%). Ideally, such lesions are intraosseous, but occasionally, they may erupt into the oral cavity.

We present a rare case of complex odontoma as it occurred bilaterally and posteriorly at the base and angle of the mandible with a large size, thus making it rare. This must be differentiated from cementoblastoma, osteoid osteoma, and fibro-osseous lesions such as cemento-ossifying fibroma. A cementoblastoma has a well-defined radiopaque mass at the root tip encircled by a radiolucent rim. Osteoid osteomas present with a small ovoid radiolucent area surrounded by a rim of sclerotic bone; the central radiolucency shows calcification. Cemento-ossifying fibroma is characterised as a well-defined radiolucency with rising speck of calcification as it matures; not bordered by a radiolucent rim and is fused with normal bone. Extended Risdon’s incision was planned to surgically excise the mass as it has less chances for facial nerve deformity, but at the same time has limitations such as reduced accessibility and damage to retromandibular vessels.
The histological picture of odontomas depicts the presence of enamel matrix, dentin, pulp tissue, and cementum that may, but need not, exhibit a normal relationship. Compound odontomas are composed of tooth-like structures that look like pulp tissue in the central portion surrounded by a dentin shell and partially covered by enamel. Complex odontomas are conglomerates without any organisation of dentin, enamel, enamel matrix, cementum, and pulp tissue. The capsule of connective tissue that surrounds an odontoma is similar to the follicle that covers a normal tooth. In our case, the histopathological section showed multiple small teeth-like structures with haphazardly arranged central fibrofatty pulpal stroma surrounded by well-formed dentin showing dentinal tubules and empty areas representing decalcified enamel matrix surrounded by fibrovascular stroma at the periphery, thus making it a complex composite odontoma.

According to Kaban, the odontomas must be surgically enucleated as it contains various tooth formulations that may predispose to cystic change and interestingly the adjacent tooth in the vicinity of odontoma is not traumatised as they are separated by a bony septum. In our case, we gently enucleated the large bony mass without much iatrogenic deformity to the shape of the jaws, thus preserving the facial aesthetics of the patient.

Hence, we recommend that an early diagnosis of odontomas is important to prevent craniofacial developmental problems. The early diagnosis accompanied by proper treatment will result in a favorable prognosis.

**Conclusion**

The takeaway lessons from this rare case report are that an early diagnosis and timely treatment of odontomas ensures minimal surrounding deformities, a better prognosis with less...
chances of recurrence, and better facial aesthetics if large in size. This case report will surely benefit many clinicians in treating complex odontomas.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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**Figure 7:** Histopathological section revealed multiple small tooth-like structures with haphazardly arranged central fibrofatty pulpal stroma surrounded by well-formed dentin showing dentinal tubules and empty areas representing decalcified enamel matrix surrounded by fibrovascular stroma at the periphery.

**Figure 8:** Facial appearance after 3 months without any jaw deformity, and radiologically intact shape of the mandible with healed fracture site on the left side body of the mandible.

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**Conflicts of interest**

There are no conflicts of interest.