Case Report

Complex odontoma: A single center case series

ABSTRACT

Odontoma is a benign mixed odontogenic tumor. Odontoma is classified into two variants: compound odontoma, which has resemblance to the tooth structure, and complex odontoma, which consists of haphazardly arranged enamel and dentin bearing no resemblance to the tooth structure. The most common site of occurrence of complex odontoma is the posterior aspect of mandible. Surgical excision of the complex odontoma remains the mainstay of treatment, which seldom recurs. Here, we report a series of complex odontoma cases reported to our institute that were managed by surgical excision with no signs of recurrence in subsequent follow-up appointments.

Keywords: Odontogenic tumors, odontomas, surgical excision

INTRODUCTION

Odontomas are grouped under the category of Benign mixed (epithelial and mesenchymal) odontogenic tumors.[1] Odontomas, previously thought to be developmental disturbances, are now considered hamartomas because of their origin in the odontogenic epithelium, and it comprises both odontogenic hard and soft tissues. Odontomas are the second most common odontogenic tumors in incidence after ameloblastoma^[2] but few consider it to be the most common odontogenic tumor as most of the cases are not documented. Odontomas are classified as complex and compound odontomas. Compound odontomas have a resemblance to the tooth structure, with the enamel and dentin arranged in an orderly fashion, whereas a complex odontoma bears no resemblance to the tooth structure, with enamel and dentin arranged in a haphazard way. Odontomas are benign and with the least aggressive potential. They can be treated with surgical excision of the tumor which seldom recurs. Here we are going to present a case series on complex odontomas reported in our institution, managed with surgical excision with no signs of recurrence in subsequent follow-up appointments.

CASE REPORTS

CASE 1

A 17-year-old male patient reported to our institutional out

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patient department (OPD) for a regular dental check-up. Upon examining the diagnostic intra oral periapical radiograph, a radiopaque mass was noticed distal to the 46. Intraoral examination revealed buccolingual expansion of the alveolar ridge distal to 46 teeth. No signs of inflammation or sinus opening were evident. Overlying mucosa is not tender upon palpation. Cone beam computed tomography (CBCT) was done, which revealed a well-defined radiopaque mass homogenous to the dentine of the adjacent tooth and surrounded by a radiolucent halo. It was extending from the distal of 46 teeth to the anterior border of the ramus anteroposteriorly and from the level of alveolar bone to the inferior alveolar canal superoinferiorly, displacing the neurovascular canal inferiorly and lingually. The distal root of the first molar showed signs of resorption mostly because of the pressure from the inferiorly

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displaced and impacted the second molar. The third molar was also impacted in an inferior and distal position to that of the radiopaque mass. The obvious radiographic picture leads to a provisional diagnosis of complex odontoma [Figure 1b]. As the patient presents no contraindication for surgery; he got operated on under general anesthesia, the tumor was surgically excised via intraoral approach, and the impacted teeth also were extracted [Figure 1a]. The biopsied specimen was sent for histopathological examination and the report came out to be as complex odontoma. Iodoform dressing was advised to promote bone fill [Figure 1c].

CASE 2

A 12-year-old female reported to our departmental outpatient department with a chief complaint of swelling since the last 1 month. She had an episode of pain with pus discharge intraorally. Upon examination, the swelling was bony hard in consistency and tender on palpation. Intraoral examination revealed swelling of 3×2 cm extending from the distal of 35teeth to the retromolar trigone of the mandible. Mucosa over swelling is normal with no evidence of active pus discharge. Radiographic detail of the lesion is in accordance with the complex odontoma, showing buccolingual cortical expansion, perforated buccal cortical plate, and an intact lingual plate of the mandible. Surgical excision of the odontoma was carried out under general anesthesia by extraoral approach performing buccal decortication and excision of the odontoma [Figure 2]. Histopathological report confirmed the diagnosis of complex odontoma.

CASE 3

A 32-year-old male came to our department with a chief

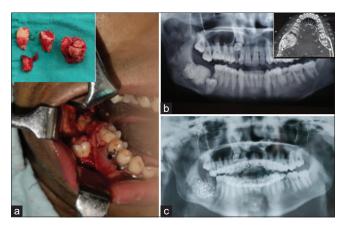


Figure 1: (a) Intraoperative picture showing the odontoma (inset – excised specimen). (b) Preoperative OPG showing the complex odontoma near the right side of mandible distal to first molar (Inset-axial section showing the buccolingual expansion of bone). (c) Postoperative OPG showing the iodoform dressing in place in the bony defect

complaint of heaviness on the right side of his face in the middle third. He was not having any swelling or area of tenderness, both extra orally and intraorally. Upon radiographic examination, a mushroom-shaped radiopaque mass was seen extending cranially from 17, occupying most of the right maxillary sinus and eroding the buccal cortical plate and anterior wall of the maxilla [Figure 3a and 3b]. Surgical excision of the mass was done through an intraoral approach, and it was excised in toto [Figure 3c and 3d]. The histopathological report confirmed the diagnosis of complex odontoma.

CASE 4

A 11-year-old male reported to our OPD with the chief complaint of swelling over the left lower third of the face in the past 4 months, which is asymptomatic otherwise. Clinical examination revealed a bony hard swelling over the left body region of mandible, which is not tender upon palpation. No lymphadenopathy was present. Intraoral examination revealed missing 37 teeth with buccolingual swelling of the left mandibular alveolar ridge distal to 36 covered with normal mucosa. Orthopantomogram (OPG) revealed an irregular radiopaque mass bounded by a well-defined radiolucent halo extending from the distal cusp of 36th tooth to the anterior border of the ramus, leaving the lower border of the mandible intact [Figure 4]. It was surgically excised in-toto via an intraoral approach, and the histopathological examination confirmed the diagnosis of complex odontoma.

CASE 5

A 9-year-old male reported to the department with a chief complaint of swelling in relation to the right side of the lower third of the face for 5 months. Upon examination, the swelling was bony hard in consistency and was non tender. Intraoral examination showed missing 47 teeth. Radiographs revealed a radiopaque mass extending from

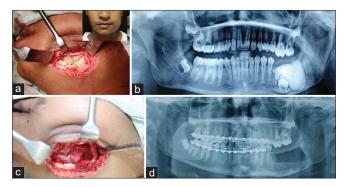


Figure 2: (a) Intraoperative image showing the odontoma (inset—preoperative clinical image). (b) OPG showing the complex odontoma near the left body of the mandible area. (c) Intraoperative image showing the intact lingual cortex after the surgical excision of odontoma (d) Postoperative OPG

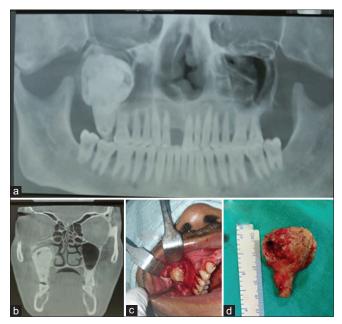


Figure 3: (a) Preoperative OPG showing odontoma near the right posterior aspect of maxilla. (b) Coronal section showing the odontoma. (c) Intraoperative image depicting the complex odontoma (d) Excised specimen

distal aspect of 46 till the mid-ramus area anteroposteriorly and from the occlusal plane to the lower border of the mandible superoinferiorly [Figure 5a]. The mass is located coronal to a displaced molar tooth, which is at the level of the inferior border of the mandible. It was surgically excised under general anesthesia. As the radiopaque mass is not well corticated and not having a plane separating it from adjacent normal bone, it was sectioned and curated out through an extraoral submandibular approach, leaving the lingual cortical plate intact [Figure 5b].

CASE 6

A 28-year-old old female reported to institutional OPD for a regular dental check-up. OPG revealed a well-defined radiopaque mass attached to the distal surface of 37 surrounded by a radiolucent Halo, extending from the occlusal level and anterior border of the ramus to the level of the inferior alveolar canal [Figure 6]. It was completely asymptomatic and an incidental finding. Surgical excision has been advised but as it was asymptomatic, she did not turn up for surgery.

DISCUSSION

Odontomas are benign mixed odontogenic tumors that occur predominantly as intraosseous variants, and there are case reports of peripheral/gingival variants of odontoma also been found in literature.^[3] Odontomas does not show any significant relationship with sex, but a few studies mentioned the female predominance compared to males.^[4] But the 4 out

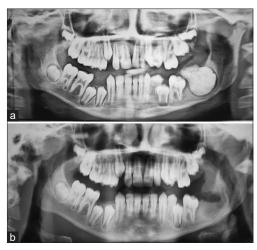


Figure 4: (a) Preoperative OPG (b) Postoperative OPG

of 6 cases presented here are in males. Odontomas are more common in permanent dentition than in primary dentition.^[5] It is classified into two variants histologically, complex and compound odontomas. Recent studies are in favor that ameloblastic fibroma is the less prominent and less organized variant of odontoma and ameloblastic fibro-dentinoma and ameloblastic fibro odontoma being the developmental stages between ameloblastic fibroma and odontoma. But molecular level analysis is still in progress to confirm this hypothesis. Compound odontoma, which resembles the tooth-like structure is more common in the maxillary anterior region. Complex odontoma, in which the dental tissues are arranged in a haphazard way, does not resemble a tooth structure and is common in the mandibular posterior region. Five of the 6 cases reported to us are in the mandibular posterior area, with one case in the maxillary posterior area. It is most common during the second and third decades of life.[6]

Etiology of the odontoma has been previously attributed to trauma, hereditary anomalies, infection of the deciduous predecessor, and epithelial remnants of cell rests of Serres. The Latest theories suggest that aberrant activation of Wnt/ β -catenin signaling may associate with odontoma formation. Most proteins involved in the differentiation of dental tissues were more immunoexpressed in compound odontomas, whereas complex odontomas show the presence of not fully differentiated cells, thus explaining the close resemblance of compound odontoma to the tooth structure, which is not in the case of complex odontoma. Ele

Odontomas can also be associated with syndromes like basal cell nevus syndrome, Gardner syndrome, familial colonic adenomatosis, Tangier disease, or Hermann syndrome.^[9]

Odontomas, most of the time, are incidental findings during routine radiologic investigations like cases 1 and 6 in our series.



Figure 5: (a) Preoperative OPG (b) Intraoperative image showing the sectioning of odontoma

Patients might complain of unerupted teeth or over-retained primary teeth. Swelling of the bony cortices and tooth displacement are more common in complex odontoma than in compound variant. Pain and lymphadenopathy can be noticed in the case of an infected odontoma. Paresthesia can also be noticed in case odontoma is compressing the neurovascular bundle.

Apart from intraosseous and peripheral variants, erupted odontoma is also reported in the literature, where the odontoma will be exposed through the oral mucosa. Possible explanations for eruption can be eruptive forces of the underlying impacted tooth or the remodeling of alveolar bone, sequestration of the overlying bone, or reactive growth of the capsule.^[10]

Conventional x-ray like OPG is sufficient to diagnose an odontoma. A CBCT can be advised to appreciate the bucco-lingual expansion and delineate the odontoma properly. Radiographically odontoma can be appreciated in three stages of gradually increasing radiopacity, ranging from the radiolucent stage, followed by the partially calcified stage, to the final, classic radiopaque stage.^[11]

Complex odontoma generally appears as a radiopaque mass with no resemblance to tooth structure and are often surrounded by a radiolucent halo corresponding to the connective tissue capsule.

Surgical excision of the odontoma remains the mainstay of the treatment, and it seldom recurs with very few cases of recurrence reported in the literature. The presence of a radiolucent halo surrounding the odontoma hints at the chances of total removal of the complex odontoma. Early stages of odontoma, which are partially calcified, necessitates sectioning of the mass to facilitate its excision.



Figure 6: Orthopantomogram showing odontoma in relation to the left angle of mandible

Blinder *et al.*^[13] emphasized the advantage of a lingual approach for excision of odontomas in the mandibular angle region. He reported a case of iatrogenic mandibular angle fracture while extracting the tooth associated with odontoma, which was then managed with the intermaxillary fixation for a period of 6 weeks. Chrcanovic *et al.* advised 2-staged surgical removal of large odontomas with an intermediate period of 3 months in between to promote bone consolidation.^[14]

Teeth impacted under the odontoma do not necessitate extraction of the teeth. Those teeth can be left to erupt spontaneously or can be managed by orthodontic interventions to facilitate their eruption. If they are severely malposed or associated with any pathologies like cyst formation, extraction of an impacted tooth is advisable.^[15]

Even in cases of defects extending through the entire height of the mandible, as in cases 2 and 5, no reconstruction has been planned considering the growing age and growth potential of the remaining intact periosteum, either in the inferior border of the mandible or intact lingual plate, which helps in spontaneous bone growth over a period of time. Temporary intermaxillary fixation was done, and patients were instructed to be cautions and not to apply any kind of force on the operated site till the healing completes.

In a few cases, to prevent the hematoma formation, packing of the surgical defect has been done using betadine-soaked ribbon gauge, whose end was left out through one end of the extraoral incision. The packing was removed on the third postoperative day. No surgical site infections or wound dehiscence were noted in any of the patients. The postoperative phase was uneventful for all the patients.

CONCLUSION

Complex odontomas, because of obvious radiological

features, can be diagnosed accurately in most of the cases. Even though they are benign and asymptomatic, they should be treated soon after diagnosing to prevent malocclusion, facial asymmetry because of expanding cortices, and other complications like infection, paresthesia and cyst formation.

Declaration of patient consent

The authors declare that they have obtained consent from patients. Patients have given their consent for their images and other clinical information to be reported in the journal. Patients understand that their names will not be published and due efforts will be made to conceal their identity but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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