Dilated odontoma: A report of two cases from a radiological perspective

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Abstract

Dilated odontoma is the most extreme form of dens invaginatus. The lesion appears as a roughly spherical mass that does not resemble a tooth but in a way appears tooth - like on radiographs due to somewhat similar radiodensity. The lesion is mostly spherical in appearance and hence the term "dilated." Occasionally, we come across cases of simultaneous pathologies. Here, we report two cases of a dilated odontoma one of which is associated with dentigerous cyst and in other case dilated odontoma pushing the maxillary sinus superiorly. Histologically, the mass was composed of dentinal tubules. These morphological and histological features are compatible with those of a dilated odontoma.

Keywords: Dens invaginatus, dens in dente, dentigerous cyst, dilated odontoma

Introduction

Dilated odontoma a variant of dens invaginatus is a developmental tooth anomaly. Morphologically, it shows wide variations that result from infolding of the enamel organ into the dental papilla before the mineralization phase.^[1] Prevalence of dilated odontoma ranges from 0.25% to 7.74%. Even though it occurs in both, the deciduous and permanent dentitions, it affects mostly the maxillary permanent incisors, particularly in the lateral incisors and very rarely the posterior teeth.^[2] Dilated odontoma currently do not feature in the most recent classifications of odontogenic tumors as an independent entity. Dilated odontoma shows a completely inverted structure of hard tissue due to the severe invagination of the enamel organ into the developing dental papilla, presenting radiographically as a shell-like structure with an outer radiopacity and a central core of radiolucency. Here, we present two extremely rare cases of dilated odontoma with a case report, treatment, follow-up, and literature review.

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Case Reports

Case 1

A 16-year-old boy reported to the Department of Oral Medicine and Radiology with swelling on the right side of the body of the mandible for the past 1 month. The swelling gradually increased in size up to a maximum size of approximately $3 \text{ cm} \times 2 \text{ cm}$. Patient revealed no contributory medical, dental, surgical, and major traumatic history. On extraoral examination, a diffuse swelling was present in the body of the mandible with ill-defined smooth borders that caused facial asymmetry. This swelling was not associated with any numbness of lower lip. The swelling was firm, fixed, nontender and nonfluctuant. On intraoral examination [Figure 1], a diffuse swelling was present in the right buccal vestibule adjacent to the right mandibular canine, premolars and the first molar obliterating the buccal vestibule. Electric pulp testing showed a delayed response in the mandibular right premolars. Mandibular occlusal radiograph [Figure 2a] revealed a well-defined radiolucent lesion with respect to the right mandibular canine, premolars and first molar with expansion and thinning of the buccal cortical plate. A radio-opaque tooth like structure was present buccally within the radiolucent lesion between the premolars. Orthopantomogram [Figure 2b] revealed a mixed radiopaque - radiolucent lesion with a hazy radiopacity surrounded by a well-defined radiolucent lesion on right side of mandible above the mental foramen. The radiopacity was seen immediately below the resorbed root of right mandibular

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first premolar along with a radio-opaque structure resembling the crown of supernumerary tooth present between the roots of the premolars. Cone beam computed tomogram [Figure 2c and d] revealed a well-defined expansile lytic lesion in the axial view, present on the right side below the level of roots which revealed a well-defined circular rim of hyperdense mass present within the outer hypodense mass. Excision was performed under local anesthesia. Excised specimen [Figure 3a] showed a hard structure present within the outer cystic mass. Histopathological section [Figure 4a] of the outer cystic mass showed a lumen, 2–4 cell layer thick epitheliums supported by dense fibrous connective tissue. Histopathological section [Figure 4b] of the hard structure showed large number of dentinal tubules.

Case 2

A 24-year-old male came with a chief complaint of occasional pain and swelling in the right upper jaw for past 8 months. The patient gave a history of a swelling and tooth pain in the same region for the past 8 months for which he underwent extraction of the upper right third molar. Intraoral



Figure 1: Case 1 intraoral picture

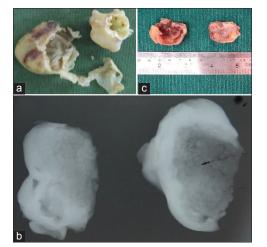


Figure 3: (a) Excised lesion in Case 1. (b) Case 2 lesional radiograph. (c) Excised lesion in Case 2

examination revealed slight swelling and erythema on the buccal aspect of maxillary right first molar and root stump of maxillary right second molar. There was no tenderness in the associated teeth. Orthopantomograph [Figure 5a] revealed a large roughly oval mixed radiopaque - radiolucent lesion in the right posterior maxillary region distal to the maxillary right second molar which seemed to be pushing the floor of the right maxillary sinus superiorly. Paranasal sinus view [Figure 5b] revealed a similar large mixed lesion in the posterior right maxilla. Coronal and axial sections of computed tomography scan [Figure 5c and 5d] revealed a well-defined hyperdense mass in right maxillary tuberosity region along the posterolateral walls of the right maxillary sinus. The lesion appeared to be donut or shell shaped with a hypodense center surrounded by a well-defined hyperdense mass. Excision was performed under general anesthesia, and radiograph of the specimen lesion was taken [Figure 3b]. Histopathological sections [Figure 4c]

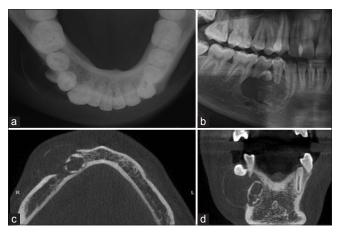


Figure 2: Case 1 (a) mandibular occlusal. (b) Cropped orthopantomograph image. (c) Axial section of cone beam computed tomogram. (d) Coronal section of cone beam computed tomogram

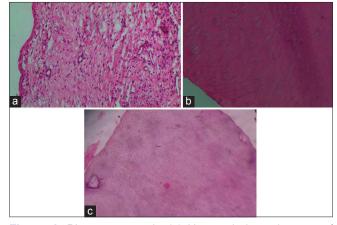


Figure 4: Photomicrograph. (a) Histopathological image of cystic mass - Case 1. (b) Histopathological image of hard tissue mass - Case 1. (c) Histopathological image of hard tissue - Case 2

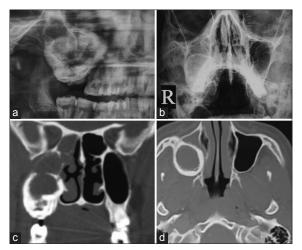


Figure 5: Case 2 (a) cropped orthopantomograph image. (b) Cropped paranasal sinus view image. (c) Coronal section of computed tomography. (d) Axial section of computed tomography

revealed large number of dentinal tubules. Based on these radiological and histopathological findings, we arrived at a diagnosis of dilated odontoma.

Discussion

A dilated odontoma present as a dilatation of the crown and root. It is considered as the most extreme variant of dens invaginatus.^[1] Dens invaginatus is an enamel lined deep invagination of the crown or root. Socrates reported the first case of dens invaginatus in human tooth. Swanson and McCarthy first presented a report of bilateral dens invaginatus.^[3] Even though the etiology of dens invaginatus malformation is debatable, various theories have been proposed to explain the etiology of dens invaginatus.^[3] Ruston described the characteristics of dilated odontoma.^[4] Dilated odontomas can be removed easily under local anesthesia. Moreover large odontoma can cause a delay in the eruption of permanent teeth and can develop cystic lesions further as dentigerous cysts.^[5] According to World Health Organization classification, odontomas can be divided into three groups.^[6]

Complex odontoma

The calcified dental tissues are simply arranged in an irregular mass bearing no morphologic similarity to rudimentary teeth.

Compound odontoma

This composed of all odontogenic tissues in an orderly pattern, which result in many teeth-like structures, but without morphologic resemblance to normal teeth.

Ameloblastic fibro-odontoma

Consist of varying amounts of calcified dental tissue and dental papilla-like tissue, the later component resembling an ameloblastic fibroma. The ameloblastic-fibro odontoma is considered as an immature precursor of complex odontoma. H M Worth described a dilated odontoma as the lesion in which the crown or root part of tooth shows marked enlargement.^[6] Oehlers classified dens invaginatus into three categories according to the depth of invagination.^[7] A dens invaginatus occurs at high incidence in the parents and/or siblings of the patients,^[1] and with other dental abnormalities such as a microdontia, macrodontia or a pumpkin-like shape with a reduced ratio of the crown and root.^[1] Therefore, genetic factors are suggested to be contributory toward the etiology. A dilated odontoma developing in the molar region of the mandible is rare. There are other types of lesions possibly categorized as a dilated odontoma in the recent literature.^[2,8,9] Crincoli *et al.*^[2] reported a case in the second molar region of the maxilla, showing a pumpkin-like morphology and a C-shaped pulp cavity. Continuity of enamel invaginated with that of the crown and pulp horn was clearly shown. Joubert et al.^[10] reported a dilated odontoma in the third molar of the mandible. In this case, the bizarre malformation in the crown and upper half of the root showed the characteristics of a complex odontoma and the lower half of the root was almost normally formed. This is an example of a dilated odontoma categorized between dens invaginatus and complex odontoma. Stavrou et al.^[9] reported a dilated odontoma in the root of the third molar of the maxilla. In this case, proliferation and ingrowth of Hertwig's epithelial root sheet may have occurred into the dental papilla at the late stage of tooth development.

Conclusion

A dilated odontoma can present itself in numerous ways as is seen in these two cases. The site was not the usual anterior maxilla as in cases of dens invaginatus; the radiological appearance may vary which further puts the clinician in a dilemma as to the diagnosis of the lesion. The findings in these two cases depict the rare variants of dilated odontoma, the first case because of association with a dentigerous cyst and the second because of its large size and location. Histological differentiation seems to be relatively well maintained in a dilated odontoma and is key to determine the correct diagnosis.

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Conflicts of interest

There are no conflicts of interest.

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