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Case Report

ATYPICAL ODONTOMAS IN SIBLINGS- A RARE ENTITY

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ABSTRACT

Odontomas are the most common benign odontogenic tumors of the oral cavity; however, their occurrence in the primary dentition is an extremely rare condition. They are considered to be developmental anomalies (hamartomas) rather than true neoplasms. We are reporting 2 siblings in which atypical large odontomas were found in the oral cavity in all four quadrants. Clinical and radiographic findings showed multiple malformed tooth-like structures of variable sizes in the posterior region of the four quadrants of the oral cavity and appearing as irregular radiopaque masses with the radiodensity with equal to tooth structure.

Key words: Odontoma, Panoramic radiograph, CBCT.

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INTRODUCTION

Odontomas are the most common odontogenic tumours of the jaws [1]. These tumours are developmental anomalies resulting from the growth of completely differentiated epithelial and mesenchymal cells that give rise to ameloblast and odontoblast [2]. In the classification of head and neck tumours by the WHO in 2005 odontomas are divided into two types: complex and compound. Multiple odontomas is characterised by numerous odontomas involving one to four quadrants of the jaw [3, 4]. The term odontomatosis and odontoma syndrome" have been used to describe multiple odontoma [5].

The purpose of the present case report is to present a rare case of multiple odontomas in the maxilla and mandible of two siblings aged 4 year old boy and 6 year old girl.

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CASE REPORT

A parent of two siblings reported to the department of Pedodontics and Preventive dentistry, PGIDS Rohtak with the chief complaint of missing teeth in both upper and lower jaws. The parents gave no history of previous extractions due to caries or trauma. The past medical and family history was unremarkable. The general physical examination of both the siblings appeared to be normal. Clinical intraoral examination of the 6 yr old girl revealed presence of central and lateral incisors in both upper and lower jaws of deciduous series. On palpation, a bulge was felt on posterior maxillary segments of both sides with an erupted odontome in the left maxillary segments (fig 1,3).Clinical examination of the patient's younger sibling who was 4 yr old boy revealed the same findings with multiple nodules present in the alveolus of posterior segment of both jaws. No other abnormality was detected intraorally. There was no extraoral swelling or cervical lymphadenopathy (fig 2,4). Radiographic examination showed multiple teeth like opacities occupying the maxilla and mandible (fig 5, fig 6). Numerous well circumscribed calcifying masses were seen mostly bordered inside by thin

radiolucent area, outside of which was a thin calcifying line. CBCT (Cone Beam Computerised Tomography) was performed to obtain more detailed information of the lesions and to assist in the planning of surgical procedures. CBCT was possible only with the older siblng as the younger one was potentially uncooperative. CBCT images revealed the extensiveness of the lesions (fig 7, fig 8 a-d). The masses occupied the alveolar part of the mandible in the body and parasymphyseal region bilaterally. Medical consultation did not confirm any syndrome. Both the siblings were found to be medically fit. Serum calcium, phosphorus, alkaline phosphatase levels were within the normal limits. According to clinical and radiographic findings, the provisional diagnosis for the older 6 yr old sibling was multiple compound odontoma whereas in case of younger sibling as the CBCT was not possible, the diagnosis was made only on the findings of panaromic radiograph. The OPG revealed multiple agglomerate masses of calcified tissues involving the alveolar and basal areas of maxilla and mandible. The lesions in the mandible involved both the parasymphyseal and body region bilaterally and in the maxilla extending from canine to molar region bilaterally. The patient was roughly diagnosed to have multiple complex odontoma.

Although the treatment plan in most of such cases involves surgical enucleation but this treatment option was not feasible in this case considering the extensiveness of the lesion. After consulting an oral surgeon it was suggested that looking into the current extent of condition any surgical procedure at this stage could compromise the growth of the jaws and could also lead to mandibular fracture since the pathology was extending to the border of the mandible. Hence the patient is kept on followup till the vertical growth of the jaws is adequate for undergoing any surgical procedure.

Fig 1. Intraoral view of the older sibling with swellings present in the alveolus, erupted four incisors in both jaws and an erupted odontome in the maxilla

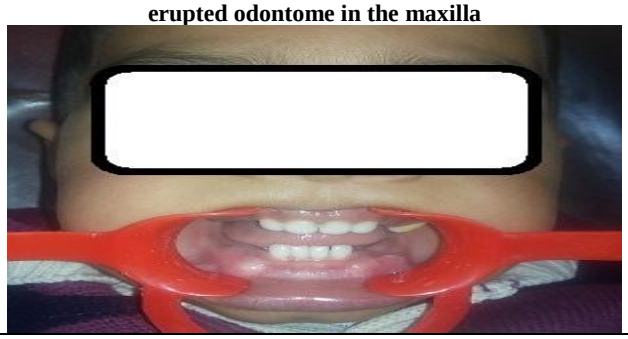


Fig 2. Extraoral view of the older sibling

Fig 3. Intraoral view of the younger siblings with multiple nodules in the alveolus and erupted incisors in both jaws

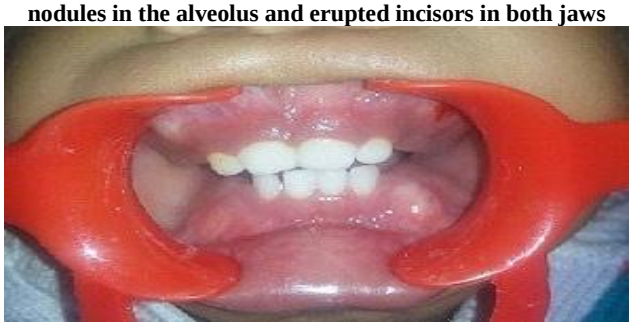


Fig 4. Extraoral view of the younger sibling

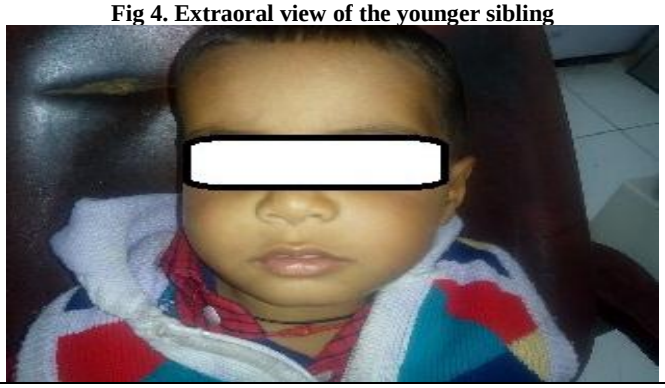


Fig 5. OPG of younger sibling showing odontomas in both the quadrants of maxilla and mandible

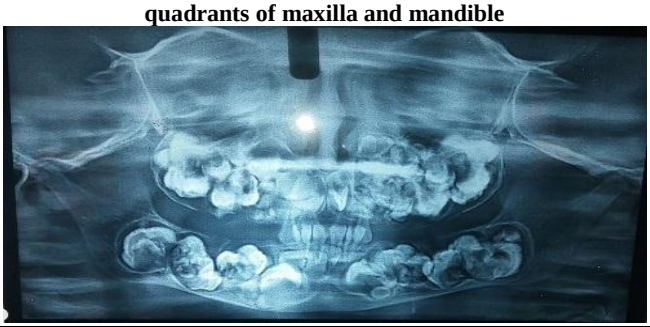


Fig 6. OPG of older sibling with one erupting odontoma on the left side of maxilla

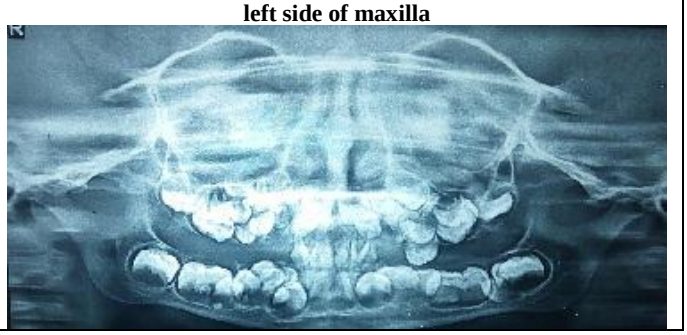


Fig 7. CBCT image of the older sibling

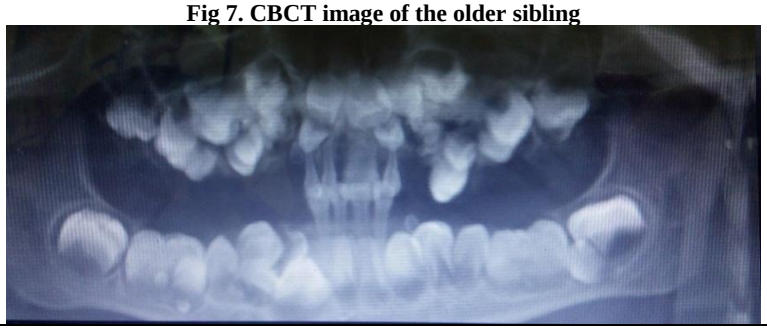
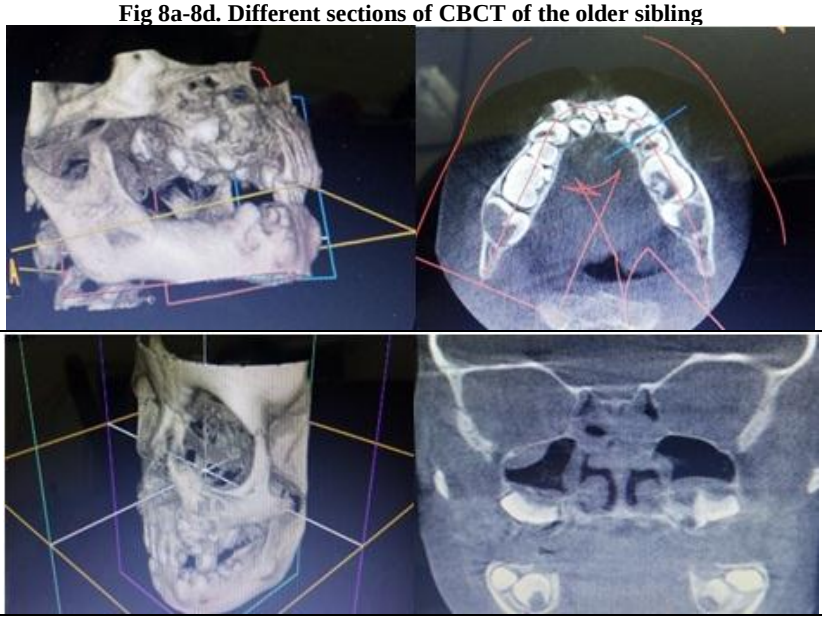


Fig 8a-8d. Different sections of CBCT of the older sibling



DISCUSSION

Although odontoma is a very common odontogenic tumour, multiple Odontomas involving numerous sites of the jaws is not frequently encountered. A review of the English language literature found only 11 such cases [6, 7]. Odontomas may be found at any age, however they are mostly detected in the first two decades of life. There is no gender predilection and most lesions are detected on routine radiographs. The maxilla is slightly more frequently affected than the mandible. Inconsistent with the usual cases, here multilple odontomas were found in the anterior as well as posterior segments of both the jaws.

The radiographic appearances of odontoma are associated with the stages of development and mineralisation with histologic differentiation to enamel and dentin. The correspondingly mixed radiopaque attenuation will appear and increase [8]. Completely radiolucent odontomas without calcification are rare. Partial calcification within a cystic radiolucency is frequently observed indicating a developing lesion. In the mature stage, the calcification occupies most of the tumour, surrounded by a narrow radiolucency. The radiographic differential diagnosis of complex compound odontomas include cememtoblastoma, ameloblastic fibro-odontoma, periapicalcementosseous dysplasia and florid cementosseous dysplasia[9].

Increasing size of odontomas may lead to sequestration of the overlying bone and produce a force sufficient to cause bone resorption, hence occlusal movement or eruption [10, 11]. Eruption of odontomas through the mucosa could also induce invasion of micro-organisms into the underlying bone due to lack of adhesion between bone and odontoma and may cause patient pain and swelling. Our patient was asymptomatic concerning pain, related neurologic and soft tissue involvement and had no aesthetic deficit due to growth of the lesions.

Follow up observations of both the siblings are ongoing. The future plan involves surgical removal of the odontomas after completion of primary growth of the jaws followed by orthodontic treatment for the remaining teeth as well as implant placement in the area of missing teeth, taking the skeletal growth into consideration. Total surgical removal of the lesion would result in massive defects, which required complicated reconstructive procedures and would also have interfered with the growth of the jaws.

CONCLUSION

We report a rare case of siblings presented with multiple large compound and complex odontomas in all four quadrants with no evidence of genetic inheritance or any signs or symptoms related to previously identified syndromes and no signs of pain or infection.

STATEMENT OF HUMAN AND ANIMAL RIGHTS

All procedures performed in human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This article does not contain any studies with animals performed by any of the authors.

CONSENT

Informed consent of the patient was obtained.

ACKNOWLEDGMENT

Nil

CONFLICT OF INTEREST

Nil

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