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CASE REPORT

A large developing complex odontome

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Abstract

Odontomas are the most common odontogenic hamartomas, broadly classified as compound or complex of which the latter is less common. Complex odontoma is a lesion in which all the dental hard tissues are represented but occur in a disorderly pattern. They are usually asymptomatic and diagnosed on radiographic examination. We report a rare case of a large developing complex odontoma in a young girl causing expansion of bone and facial asymmetry with the failure of eruption of several mandibular posterior teeth. The odontoma was treated by careful surgical curettage, marginal mandibular rim resection and bone grafting. Follow-up over a couple of years revealed no evidence of recurrence.

Introduction

Odontoma is a benign odontogenic hamartoma, constituting 22% of all odontogenic tumours[1]. Based on radiographic and microscopic features, odontomas are classified into complex and compound subtypes. Complex odontomas are defined as malformations in which all of the dental tissues are represented, and individual tissues are well formed but occur in a disorderly pattern[2]. They tend to occur in the posterior mandible presenting in the second decade of life with a slight female preponderance[3]. Complex odontomas can be associated with pathologic changes such as impaction, malpositioning, aplasia, malformation and devitalisation of adjacent teeth[4] and are usually diagnosed on routine radiological examination. The aetiology of complex odontomas is unknown but theories include local trauma, infection, family history and genetic mutation[3]. Management is either by serial monitoring or surgical removal followed by histological analysis. We report an unusually large expansile 'Developing complex odontome' in the posterior left mandible of a young girl with associated facial asymmetry and unerupted permanent dentition.

Case report

A 13-year-old Caucasian girl was initially seen by her dentist and referred to the Oral and Maxillofacial Surgery department at 'Great Ormand Street Children's Hospital' with missing left mandibular teeth. Extra-oral examination revealed slight facial asymmetry with enlargement of the left mandibular angle region. Intraoral examination displayed an asymptomatic hard, non-tender swelling in the left mandible with several missing left mandibular permanent teeth and overlying normal mucosa. The patient had no history of trauma or infections.

Initial Dental Panoramic Tomography (DPT) (Fig. 1) revealed a poorly-defined, mixed density lesion in the left posterior mandible. Associated were the displaced unerupted permanent left canine, both premolars and the first molar teeth. CT scan (Fig. 2) showed the expansion of the buccal and lingual cortical plates, with the lesion occupying a zone from the midline to the angle of the left mandible with an ill-defined transition zone. The largest area of soft tissue was on the alveolar surface of the mandible in

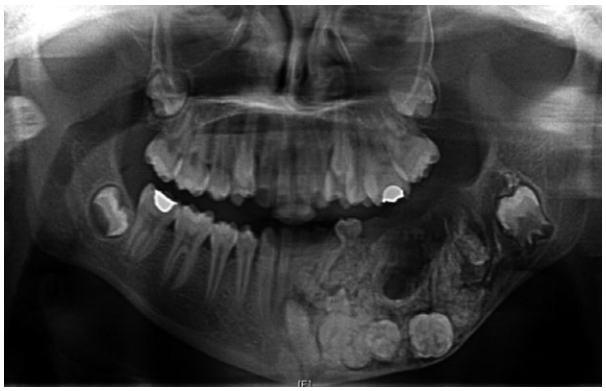

Figure 1 DPT revealing mixed radiolucent/radiopaque lesion in left mandible.

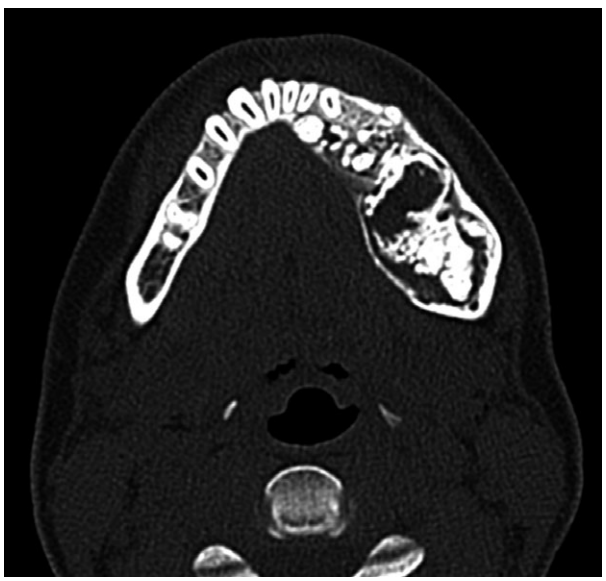

Figure 2 Axial CT scan section showing the extent of the lesion anteroposteriorly.

the molar/premolar region corresponding to the large radiolucent lytic defect on the DPT. The border of the mandible was well circumscribed except distal to the canine, where the margin was ill-defined.

A provisional diagnosis of 'Developing complex odontome' was made due to the degree of radiolucency present and partial calcification of odontogenic tissue. 'Ameloblastic fibro-odontome' and 'Odontoameloblastoma' were considered in the differential diagnosis.

Biopsy of the lesion suggested a 'Developing complex odontome'. MDT decision was to carefully remove the lesion under general anaesthetic. Removal also involved a left mandibular marginal rim resection, which was then secured with a plate. The defect was filled with bone harvested from the left iliac crest. The lower left third molar was maintained with possible future eruption potential. The specimen was sent for further histopathological examination. The patient declined immediate replacement of the lower left mandibular teeth. In the future, further bone may need to be grafted depending on the type of long-term tooth prosthetic replacement.

Histology sections of the mandibular mass showed a lobulated lesion, comprising interconnected sheets of tubular and dysplastic dentine within which were several spaces containing remnants of basophilic enamel matrix, juxtaposed to reduced enamel epithelium. Dentine was surrounded by mesenchymal tissue resembling dental papilla. The latter was abundant in the central parts of the lesion, where it featured frequent islands of odontogenic epithelium comprising peripheral ameloblast-like cells and central stellate reticulum-like cells. There were no mitotic figures or any other atypical features. Figure 3 (a and b) show histology sections. Taking into consideration the clinical, radiographic and histological find-

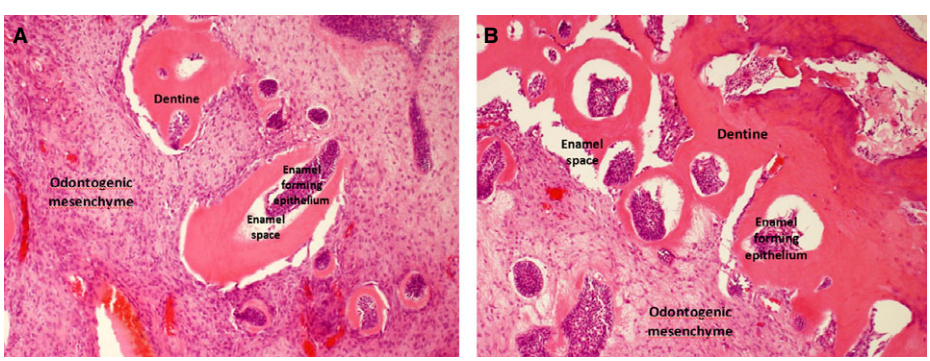

Figure 3 (a and b) Histological slides of the developing complex odontome.

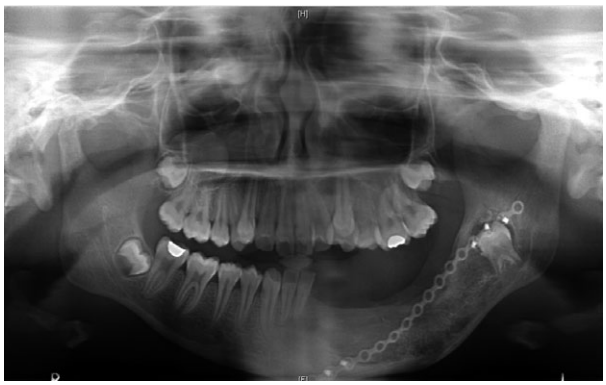

Figure 4 Six-month post-operative DPT.

ings, a definitive diagnosis of a 'Developing complex odontome' was made.

Immediate post-operative intraoral healing was good without any defects and signs of infection of the graft and donor sites. There was slight residual paraesthesia of the areas supplied by inferior alveolar nerve on the left side which was improving with time. Six-month post-operative DPT (Fig. 4) revealed good bony healing without any recurrence.

Discussion

Large 'Developing complex odontomas' are rare. Histological differential diagnosis includes 'Developing complex odontome' and 'Ameloblastic fibro-odontome'. The latter is difficult to differentiate from the immature phase of a 'Developing odontome' when hard tissue formation is minimal. However, in this case, the lesion showed abundant hard tissue arranged as a central mass with radiating layers of dentine and immature enamel. The soft tissue component at the periphery comprised dental papilla-like tissue with enamel organ-like epithelium; features not compatible with 'Ameloblastomas'. As 'Ameloblastoma'- like areas were not seen, the rare 'Odonto-ameloblastoma' was also ruled out.

Early detection and investigation of delayed eruption of teeth by radiographic examination would have helped in early diagnosis. This may have allowed for more conservative management and reduction of damage to local structures. Monitoring at this stage may have resulted in a pathological fracture of the mandible. Surgical intervention in this case is unlikely to have significant affect on mandibular growth with condylar growth and surface remodelling is still possible.

Recurrence is rare and usually only occurs in cases of incompletely excised immature odontomes[5].

Conclusion

We have reported a case of a large 'Developing complex odontome' that presented as a painless swelling in the left mandible with associated unerupted permanent teeth. The lesion was treated by surgical excision, marginal mandibular rim resection and bone grafting from the ileac crest. There was no sign of recurrence. Early detection is key for diagnosis and treatment and can help reduce damage to local structures and patient dehabilitation.

Acknowledgement

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Conflict of Interest

The authors confirm that there are no conflicts of interest.

Ethical Approval

None required.

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