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Odontogenic myxoma in the paediatric patient: a review of eight cases

S. S. Subramaniam, A. A. Heggie, R. Kumar, J. M. Shand
Department of Oral and Maxillofacial Surgery, Royal Children's Hospital of Melbourne, Melbourne, Victoria, Australia

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Abstract. Paediatric odontogenic myxoma (OM) is a rare pathological condition in the oral and maxillofacial region. There has been much debate in the literature regarding the preferred method of treatment; however due to the rare nature of this disease, definitive algorithms of management are yet to be determined. A case series of eight paediatric patients with OM is presented. Six of the lesions were in the maxilla and two were mandibular lesions. The patients were aged between 2 and 18 years. Treatment ranged from excision and the application of Carnoy's solution to segmental resection and reconstruction. From this case series it can be seen that even in situations where treatment was limited to excision and the application of Carnoy's solution, no recurrences occurred. As such the present authors favour an initially more conservative approach to the management of these lesions where possible and reserving conventional resective treatment for recurrences, lesions causing pathological fracture, and those in regions that are difficult to access.

Key words: odontogenic myxoma; paediatric.

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Since first described in 1947 by Thoma and Goldman,[1] odontogenic myxoma (OM) has remained a management dilemma for oral and maxillofacial surgeons. It is a rare, benign odontogenic tumour of mesenchymal origin that is not encapsulated and exhibits variable clinical behaviour with differences in the reported rate of recurrence.[2,3]

Clinically, the OM is often asymptomatic or presents as a firm expansile mass in the maxillofacial complex, with or without displacement or mobility of the associated dentition. There is an equal distribution between the maxilla and mandible.[4] Its incidence is approximately 0.07 per million people and it makes up approximately 0.5–20% of odontogenic tumours in adults[2,5–7] and 8.5–11.6% in children.[2,8]

Various terms have been used to describe the radiological features of OM, such as 'soap bubble' and 'honeycombing', yet none are pathognomonic.[9] Lesions may be unilocular or multilocular and may be either well-defined or poorly demarcated. These lesions can cause expansion of the bony cortex and displacement or resorption of associated teeth.[3,4,7] Histopathological features include spindle-, wedge-, or stellate-shaped cells that are loosely arranged in an abundant mucoid background.[3,7]

There is no consensus as to the mechanism of tumour infiltration, although several hypotheses have been suggested. These include the expression of matrix metalloproteinases, genetic alterations, the expression of anti-apoptotic proteins, and alterations in receptor activator of nuclear factor kappa B ligand (RANKL), its receptor, and osteoprotegerin.[7]

Due to the relatively low incidence of OM and its variable clinicopathological behaviour, it has been difficult to establish a definitive management algorithm and

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there is no widely accepted consensus. Current recommendations for treatment vary from enucleation to surgical resection with 1-cm margins. In the paediatric population, if a more radical surgical intervention is advocated, consideration must be given to the impact on growth and development, as well as impairment secondary to the ablation of vital structures.

The aim of this retrospective study was to review cases of OM in children treated at a children's hospital in Melbourne, Australia between February 2004 and February 2016 and to evaluate the mode of presentation, radiological findings, and outcomes of treatment. Approval for this study was obtained from the necessary ethics committee.

Materials and methods

A retrospective analysis of the oral and maxillofacial surgery database was performed by manual and electronic search using Microsoft Excel. This search yielded eight cases seen over a 12-year period. The inclusion criterion was any child treated for OM at the hospital; no exclusion criteria were applied to the dataset. The age and mode of presentation, radiological findings, operative approaches, and outcomes during the follow-up period were extracted from the patient records and analysed.

Results

Clinical presentation and diagnosis

In this series, the age at presentation ranged from 2 to 18 years (Table 1). Maxillary lesions were more common in the younger age group, with all cases except one being $\leq$11 years of age; these presented as asymptomatic, firm, painless swellings over the maxilla. In contrast, mandibular lesions in this series occurred in older patients and were incidental radiological findings during routine orthodontic or dental care. There were no associated signs or symptoms such as pain, mobility of teeth, or paresthesia.

In six of the cases, incisional biopsies were performed as the first diagnostic step prior to definitive treatment. In one patient (patient 2), a presumptive diagnosis of OM was made and confirmed by an intraoperative frozen section. This lesion was then treated by excision, together with a peripheral ostectomy and the application of Carnoy's solution. In another case (patient 6), the patient underwent an excisional biopsy with subsequent resection of the lesion following histopathological confirmation.

Radiological findings

All lesions were imaged using panoramic radiographs together with either a

Table 1. Data for eight patients with odontogenic myxoma treated at the Royal Children's Hospital Melbourne.

Patient	Age (years)	Presentation	Imaging	Treatment	Follow-up (months)	Recurrence
1	2	Painless firm swelling in the right anterior maxilla	CT: well-defined, expansile cystic lesion in the right maxilla/paranasal area	Right partial maxillectomy and costochondral rib graft reconstruction	103	Nil
2	2	Swelling in the right infra-orbital area with epiphora	CT: well-defined expansile cystic lesion in the right anterior maxilla/paranasal area	Excision, peripheral ostectomy, and application of Carnoy's solution	44	Nil
3	2	Left nasomaxillary facial swelling	CT: multiloculated radiolucency with bony expansion in the left paranasal area causing displacement of the developing dentition	Enucleation, curettage, and Carnoy's solution	10	Nil
4	11	Swelling over the right maxilla	DPR and CT: well-defined, expansile mass with multiple internal septations in the right posterior maxilla	Right partial maxillectomy and DCIA reconstruction	30	Nil
5	13	Painless swelling in the left maxillary sulcus, with mobile 27	DPR: left maxillary radio-opacity with displaced 28 CT: well-defined mass occupying the left maxillary sinus, extending to the orbital floor and pterygoid plates	Left sub-total maxillectomy with intraoral and infratemporal approach and DCIA reconstruction	132	Nil
6	16	Incidental finding of unilocular radiolucency between 35/36	DPR/CBCT: unilocular radiolucency between 35/36 without displacement	Marginal resection with immediate bone grafting	7	Nil
7	16	Mobile symptomatic deciduous tooth in the left maxilla	DPR and CT: Ill-defined radio-opacity involving the left maxillary sinus	Left subtotal maxillectomy and DCIA reconstruction with temporal artery anastomosis	48	Nil
8	18	Painless swelling in the left mandible	DPR: unilocular radiolucency of the left mandible involving 34/35/37/38 CT: expansile, lytic lesion in the left mandible with cortical breech and soft tissue extension	Enucleation, removal of 34/35/37/38 with Carnoy's solution	12	Nil

CBCT, cone beam computed tomography; CT, computed tomography; DCIA, deep circumflex iliac artery flap; DPR, panoramic radiograph.