

CASE REPORT

Gingival myofibroma in children: report of 4 cases with immunohistochemical findings

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Abstract

Oral myofibroma is a rare tumour which usually occurs in children and has been reported in the mandible, tongue, buccal mucosa with only a few cases reported from the gingiva. It appears alarming clinically due to its fast growth which may mimic a malignancy. However, it is completely benign and is usually treated by complete surgical excision with excellent prognosis. Clinically, myofibroma presents as a single swelling when it occurs on the gingiva, and more common lesions such as fibrous epulis, pyogenic granuloma and peripheral odontogenic fibroma, myofibroma are usually considered in the differential diagnosis. We present 4 additional cases of gingival myofibroma in children. Their ages ranged from 7 to 14 years. Three were girls and 1 patient was a boy. All presented with solitary gingival growths, ranging from 3 weeks to 2 months in duration, and raised the clinical diagnoses of peripheral giant cell granuloma, pyogenic granuloma and fibrous epulis. Histopathology of incisional biopsies revealed proliferation of streaming and whorled fascicles of spindle cells around slit-like vascular spaces. The spindle cells were cytologically bland and were immunopositive for vimentin and smooth muscle actin, but were negative for desmin and S-100 protein. All were treated by surgical excision.

Key words: oral myofibroma, gingiva, immunohistochemistry

INTRODUCTION

Myofibroma is an uncommon tumour with a predilection for the head and neck, followed by the trunk and extremities.¹ Oral myofibromas have been reported occurring in the mandible, tongue, buccal mucosa, hard palate, floor of mouth and gingiva.²⁻⁹ So far 5 cases of myofibroma of gingival location have been reported in the English language literature with 2 cases reported in children.^{4,7-9} The aim of this article is to report an additional 4 cases of gingival myofibroma in children and briefly discussed the clinical, histopathological and immunohistochemical features of oral myofibroma.

CASE REPORTS

Case 1

An 8-year-old Malay girl presented with a fast growing swelling over the lower left first molar area which was noticed for about 3 weeks. Intra-oral examination revealed a reddish, multilobulated growth at the gum of the molar

tooth which bled profusely on probing. The size of the swelling was 3.0 x 2.5 cm. The tooth was mobile. Radiography revealed loss of the coronal half of alveolar bone around the region. The clinical differential diagnosis was peripheral giant cell granuloma. Incisional biopsy was done and the specimen was sent for histopathological evaluation. A diagnosis of myofibroma was given. Excision of the lesion was then done under general anaesthesia to remove the lesion.

Case 2

A 14-year-old Malay girl had a painless growth of unknown duration in the lower right gum. On examination, an exuberant growth was noted on the gum adjacent to the lower right third molar. The growth had extended to the second molar buccally and lingually. The clinical differential diagnoses were pyogenic granuloma and malignant tumour. An incisional biopsy was diagnosed as myofibroma. Excisional biopsy using electrocautery was then performed under general anaesthesia.

Case 3

A 7-year-old Malay boy presented with a gum swelling of the lower left jaw. According to his mother, the swelling was first noticed 2 months previously and was increasing in size. However it was painless. Intra-oral examination revealed a large firm growth measuring 3 x 2 cm in size on the lingual aspect of the lower left second deciduous molar and first permanent molar. The growth extended to the distal gingiva of the permanent molar. The tooth was mobile. The growth was of normal mucosal colour and slightly rubbery in texture. Radiographic examination showed no bony changes. The clinical diagnosis was fibrous epulis. Excisional biopsy was performed and the tissue was sent for histopathological examination.

Case 4

A 7-year-old Chinese girl presented with an asymptomatic gum swelling on the lower left jaw of 2-month duration. The lesion had caused displacement on the lower left deciduous canine, first and second molars. Mucosal surface ulceration was noted. The clinical diagnosis was pyogenic granuloma. Excisional biopsy was performed and the specimen was sent for routine histopathological assessment.

Histopathology

All the incisional and excisional specimens were fixed in 10% formalin and routinely processed for light microscopical examination. Immunohistochemistry using the standard avidin-biotin peroxidase complex method was performed with antibodies to vimentin, smooth muscle actin, desmin and S-100 protein together with positive controls. Microscopically all the cases revealed similar histopathological features. The sections showed proliferation of streaming or whorled fascicles of spindle cells around slit-like vascular spaces (Figure 1). The spindle cells were cytologically bland and were immunopositive for vimentin and smooth muscle actin, but were negative for desmin and S-100 protein.

DISCUSSION

In 1981, Chung & Enzinger reported 61 cases of solitary and multicentric types of infantile myofibromatosis occurring in infants and young children.¹ Later, five cases of myofibromatosis in adults were reported.¹⁰ According to the 2002 WHO classification of soft tissue tumours, myofibroma and myofibromatosis are terms used to denote the solitary (myofibroma) or multicentric (myofibromatosis) occurrence of benign neoplasms composed of contractile

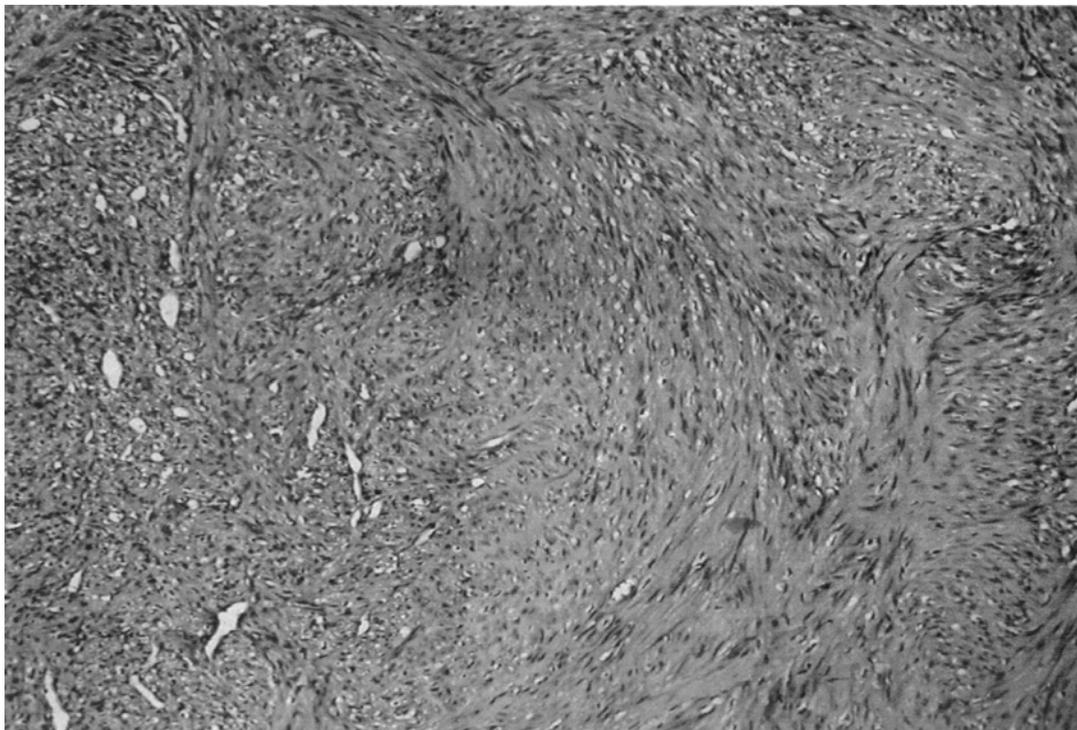


FIG 1: Proliferation of streaming and whorled fascicles of spindle cells around slit-like vascular spaces.

myoid cells arranged around thin-walled blood vessels.¹¹

Oral myofibromas are usually found in children.⁵ In a recent review done by Vered *et al*, out of 44 cases of oral myofibroma reported in the literature, 22 cases were in children below the age of 16 years old.³ In a series of 79 oral myofibromas reported by Foss *et al*, 23 cases were in the first decade and 13 cases occurred in the second decade of life.⁵ In the present study, all the patients were young ranging in age from 7 to 14 years.

Clinically, a myofibroma presents as palpable rubbery firm to hard mass either superficially located or as a fixed nodule which is deeply seated. When the skin is involved, the lesion may manifest as a purplish macule.¹ An oral myofibroma usually appears as a painless mass with rapid enlargement and secondary ulceration. When the gingiva is affected by the lesion, it presents as a swelling, hence the clinical differential diagnosis of pyogenic granuloma and fibrous epulis are favoured by the clinician because of the tendency of these two lesions to occur at the gingiva. To date, there are 5 reported cases of gingival myofibroma in the English language literature.^{4,7,9} Only 2 cases were reported in children.⁷⁻⁸ Table 1 shows the details of 9 cases of gingival myofibroma with 6 cases, including the current 4 cases, occurring in children.

The cause of myofibroma is presently unknown. Trauma or injury may contribute to

the development of the lesion and it is believed that myofibroma is derived from myofibroblast cells. Myofibroblasts are thought to play a role in wound healing.¹² Myofibroblasts are characterized morphologically as spindle-shaped cells with ill-defined eosinophilic cytoplasm and nuclei that are either symmetrically tapered and wavy or rather plump, more rounded and vesicular with indentations and small nucleoli.¹³ The myofibroblast is ultrastructurally a highly differentiated cell with specialized organelles having features in common with smooth muscle cells and fibroblast.¹⁴

Macroscopically, myofibromas are fairly well-circumscribed, unencapsulated masses. Histologically, the lesion has a whorled or nodular appearance. The nodules are composed of cells intermediate in appearance between fibroblast-like spindle cells and plump fusiform cell resembling smooth muscle cells arranged in short bundles or fascicles. The plump spindle-shaped cells contain oval to elongated nuclei with rounded or blunt ends, and have pale eosinophilic cytoplasm with fairly well-defined cytoplasmic membrane. A prominent vascular pattern with multiple thin-walled, slit like vascular spaces which are centrally located and usually resembling hemangiopericytoma is present. In these areas, the tumour vessels are closely associated with packed or loosely arranged round to polyhedral cells having large slightly pleomorphic, hyperchromatic nuclei and relatively scanty, pale pink, poorly outlined cytoplasm.¹

TABLE 1: Reported cases of gingival myofibroma in adults and children

Authors	Age	Gender	Clinical presentation	Size (cm)	Duration	Initial diagnosis
Beham et al ⁹	60 yrs	M	swelling	0.5	NI	NI
Jones et al ⁷	70 yrs	F	swelling	0.8x 0.5	4 months	Atypical cellular smooth muscle tumour
Jones et al ⁷	8 yrs	F	swelling	1.5 x 0.7	2 weeks	Myofibromatosis
de Souza et al ⁸	9 yrs	F	swelling	2.0	3 months	Peripheral ossifying fibroma Peripheral giant cell granuloma neurofibroma
Montgomery et al ⁴	50 yrs	M	swelling	2.2	NI	NI
Case 1	8 yrs	F	swelling	3.0 x 2.5	3 weeks	Peripheral giant cell granuloma
Case 2	14 yrs	F	swelling	3.5 x 2.0	NA	Pyogenic granuloma ? malignant tumour
Case 3	7 yrs	M	swelling	3.0 x 2.0	2 months	NA
Case 4	7 yrs	F	swelling	2.5 x 2.0	2 months	Pyogenic granuloma

F: female, M: male

NI: not indicated, NA: not available

Immunohistochemical investigations are usually used to support the histological diagnosis. The more eosinophilic spindle cell areas are more likely to demonstrate staining for muscle specific actin and vimentin, but are negative for desmin and S-100 protein.¹⁵ The results in this study support these findings.

Oral myofibroma usually appears alarming to the clinician because of its rapid growth which may resemble a malignancy. It is however completely benign and upon complete surgical excision has an excellent prognosis. Biopsy is mandatory for accurate diagnosis and to prevent a more radical and aggressive treatment being instated. The recurrence rate is low if the lesion is properly treated. de Souza *et al* reported a case of a gingival myofibroma in a 9-year-old girl with no signs of recurrence observed after a 2-year clinical and radiographical follow-up.⁸

Myofibroma should be considered in the management of a solitary gingival growth other than the more common differential diagnoses such as fibrous epulis, pyogenic granuloma and peripheral odontogenic fibroma.

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