Peripheral odontogenic myxoma

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ABSTRACT

Odontogenic myxomas are a rare benign odontogenic mesenchymal tumor found exclusively in the tooth-bearing area of the jaw and are usually located centrally in the mandible. Soft tissue localization is rarely seen and is classified as peripheral odontogenic myxoma (POM). POM is slow growing and less aggressive as compared to central myxoma. It has a low recurrence rate, comprises 3–6% of all odontogenic tumors. Only a few cases of POM on maxillary gingiva are reported in the literature. Here, we present an unusual case of primary POM occurring in the gingiva of anterior maxilla in a 14-year-old female patient.

Key words: Maxilla, odontogenic myxoma, peripheral

CASE REPORT

A 14-year-old female reported to the Department of Oral and Maxillofacial surgery with the complaint of swelling in the upper right front tooth region since 3 months. She underwent excision of the same type of swelling 1 year back which was of sudden onset and same size and at the same site and which was diagnosed as pyogenic granuloma on the basis of histopathological examination. On clinical examination, a single swelling on the buccal gingiva in relation to 12 and 13 was seen measuring 2 X 2 cm anteroposteriorly and mediolaterally. Swelling was oval, reddish-pink, sessile with smooth and shiny surface [Figure 1]. On palpation, swelling was nontender, soft to firm in consistency, noncompressible, and fixed to the underlying mucosa.

A preoperative intraoral periapical radiograph revealed no radiographic changes [Figure 2]. On the basis of clinical and radiographic examination, the differential diagnoses were peripheral giant cell granuloma, pyogenic granuloma, and peripheral ossifying fibroma. Surgical excision of the lesion was done. Surgical procedure was performed under local anesthesia. Excisional biopsy and curettage were done followed by peripheral ostectomy with roseheaded acrylic trimming bur, and the defect was primarily closed with partial thickness flap beyond the mucogingival junction to facilitate for coronal advancing of the flap in relation to lateral incisor and canine [Figure 3]. Excised specimen was sent for histopathological examination.

Macroscopically, tumor mass was pedunculated, pinkish-red with soft rubbery consistency, measuring approximately...
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1.5 cm² in overall dimensions. The cut surface showed jelly-like whitish appearance.

On microscopic examination, H and E stained section showed parakeratinized stratified squamous epithelium with subepithelial dense fibrous connective tissue capsule. Deeper areas showed large loosely arranged myxoid areas with the presence of stellate, angular, and spindle-shaped oval fibroblasts [Figure 4a]. Focal areas showed odontogenic rests. Delicate and thin blood vessels were evident in the myxoid stroma. Application of special stain, i.e. alcian blue at low pH was done, myxoid intercellular matrix and loosely arranged collagen fibrils showed positivity for alcian blue [Figure 4b]. Further alcian blue with periodic acid–Schiff (PAS) staining was done. Stroma showed negativity for PAS. Based on H and E and alcian blue with PAS stain, a final diagnosis of POM was given.

DISCUSSION

Odontogenic myxoma is commonly reported as slow growing usually asymptomatic, although some patients experience progressive pain in lesions involving maxilla and maxillary sinus, with neurological disturbances. Central type of myxoma has a tendency of bone destruction with high recurrence rate. Our case of the peripheral type of myxoma was slow growing and localized in nature. These tumors show a different type of radiographic feature such as small unilocular or large multilocular lesions, which usually also show displacement of teeth followed with root resorption. Various descriptions given for the multilocular pattern are honeycomb, soap bubble, tennis racket, wispy and spider-web appearance. In our case, tumor was peripherally located in gingiva hence no radiographic changes were evident.

In the international histological classification of odontogenic tumors, odontogenic myxoma is defined as a benign odontogenic tumor of mesenchymal origin that is locally invasive and consists of round to angular cells that lie in the abundant mucoid stroma.
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POM may be difficult to differentiate microscopically from other tumors with myxoid features, many of which have malignant characteristics. Their low prevalence in gingival soft tissue contributes to POMs being generally excluded from its histopathological differential diagnoses of extra bony myxoid proliferations such as soft tissue myxoma, myxomatous degeneration in fibrous lesion, focal mucinosis, nerve sheath myxoma, myxoid neurofibroma, and myxoid chondrosarcoma.\[11\]

The neoplasm is composed of haphazardly arranged stellate, spindle-shaped, and round cells in a loose myxoid stroma as seen in our case. Although the nuclei of the cells may show atypical features, mitoses are rarely seen. The production of abundant myxoid ground substance that stains positive for glycosaminoglycans, hyaluronic acid, and chondroitin sulfate is the histochemical hallmark of the neoplasm. Present case showed positivity for alcian blue stain demonstrating the presence of hyaluronic acid and chondroitin sulfate. Stromal component shows negativity for PAS demonstrating absence of neutral mucins. The presence of inactive odontogenic epithelial remnants are not required for the diagnosis, but they were identified in our case.\[12\]

Since its original description, the nature of odontogenic myxoma has been a matter of controversy. The origin of odontogenic myxomas from odontogenic ectomesenchyme was put forth by Adekeye et al. in 1984. The author suggested that rarity of odontogenic myxoma in any extragnathic bone could be the firm reason for suggesting the odontogenic origin.\[13\]

One of the hypotheses of origin is altered/primitive fibroblasts or myofibroblasts producing excess of mucopolysaccharides and was commonly incapable of forming mature collagen even if some cells could retain this capacity.\[14\] Even their origin from periodontal tissue has also been suggested.\[9\]

Ultrastructural and immunohistological studies carried out on odontogenic myxoma stress on the histogenesis and characteristics of the cell and matrix in tumor mass. A study conducted by Martínez-Mata et al. in 2008 shows that odontogenic myxoma frequently shows myofibroblast and mast cell, their presence may induce modification in the extracellular matrix and could contribute to the tumor invasion.\[15\]

Literature review conducted by Jaeger et al. in 2000 report the following findings. (1) the tumor cells are secretary, (2) the cells are mesenchymal and express vimentin, (3) positivity to S-100 protein and muscle-specific actin has been variable, (4) the matrix exhibits different protein, mostly collagen Type I and Type VI, tenascin, fibronectin, and proteoglycans.\[16\]

A study conducted by Martínez-Mata et al. in 2008 on 62 cases of odontogenic myxoma show variable expression of different markers. Epithelial islands show erratic positivity for CK 14, CK5, CK7, CK19. Stellate and spindle-shaped cells were positive for vimentin while a few cases were positive for α-AML, HHF35, S-100 and neuron-specific enolase. However, further studies will help to elucidate its behavior.\[15\]

Odontogenic myxoma shows locally aggressive feature with high recurrence rate; hence, the treatment of choice is radical resection. However, soft tissue myxoma is not reported to cause recurrence or metastasis;\[17\] hence, carefully planned conservative treatment or semi-radical approach is justified in these cases. In this case, the tumor was completely removed by local surgical excision and there was no recurrence after 9 months of surgery [Figure 5].

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Conflicts of interest
There are no conflicts of interest.

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Figure 5: No recurrence after 9 months of follow-up
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