

Odontogenic Fibromyxoma of the Maxilla: A Rare Clinical Entity

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Abstract

Odontogenic fibromyxoma represents a rare benign odontogenic tumor of mesenchymal origin which exhibits locally aggressive behavior and is prone to local recurrence, affecting mostly the posterior region of the mandible. In this paper, we present a case report of a 25-year-old lactating female with the gingival swelling of the hard palate. The swelling was initially localized to the gingiva and was small in size but had increased in size over a year. The patient had discomfort while eating as she was biting on the mass during mastication. The clinical findings matched with the final diagnosis of odontogenic fibromyxoma which was established through subsequent histopathological examination. Odontogenic fibromyxoma is a diagnostic and therapeutic challenge. Hence, the aggressiveness and the high potential of the maxillary fibromyxoma recurrence suggest that a wide surgical resection is the best treatment option to guarantee a good prognosis. The odontogenic fibromyxoma, though not life threatening in the immediate time span, is annoyingly symptomatic and affects the quality of life badly because of its location and nature. Hence, complete removal of the tumor including the involved tooth, adjacent soft tissue, and bone is advised.

Keywords: Fibromyxoma, Maxilla, Odontogenic tumor

INTRODUCTION

Odontogenic fibromyxoma is synonymous with odontogenic myxoma, according to the World Health Organization. It represents 0.04–0.06% of oral cavity tumors and 3–7% of benign odontogenic tumors.^[1] It usually occurs more in women aged 20–30 years old than in males.^[2] Fibromyxomas in the head-and-neck region commonly originate either from the soft tissues or from the facial skeleton. Although it is believed to derive from embryonic mesenchymal elements of the dental papilla, dental follicle, or periodontal ligament,^[3] still, the etiology of fibromyxomas remains indeterminable.

The clinical presentation of fibromyxoma is varied. Majority of patients with odontogenic fibromyxomas present with a painless swelling.^[4] It is usually a benign, slow growing, and rare odontogenic tumor, which is locally invasive of the maxillary ectomesenchyme with or without epithelial induction. Radiological images are not specific enough and only histopathological examinations can confirm the diagnosis.

Surgical removal is the only treatment of choice.

Thus, the aim of this particular paper is to present the rarity of a fibromyxoma of the maxilla, the contribution of the radiological examination to the differential diagnosis, and the importance of a meticulous excision to prevent recurrence.

CASE PRESENTATION

A 25-year-old lactating female reported with a gingival swelling on the hard palate. The swelling was initially localized to the gingiva and was small in size but had increased in size over a year. The patient had discomfort while eating as she was biting on the mass during mastication. However, she denied any history of fever, chills, night sweats, or weight loss. There was no facial or cervical lymphadenopathy or any associated pain.

On intraoral inspection, a reddish smooth surface growth of 4 cm × 5 cm in greatest dimension was noted, which was diffuse and non-proliferative, pedunculated, non-ulcerative, non-hemorrhagic, firm in consistency, non-pulsatile in nature (Figure 1). The mass extended from the left palatal area adjacent to the molars to 1 cm medial to the left alveolar margin and anteroposteriorly from the level of the left canine to 5 cm posteriorly without causing any displacement of any molar or premolar teeth.

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Cone beam computed tomography (axial section) of the left maxilla revealed radiolucency in the palatal root of upper left first molar involving the furcation area (Figure 2). Pre-operative routine blood investigation was within normal range and reverse transcription polymerase chain reaction for SARS-COV-2 was found negative.

An incisional biopsy was performed under local anesthesia and was sent for the histopathological diagnosis.

The histopathological examination revealed thin normal stratified squamous epithelium supported by loose gelatinous myxomatous connective tissue stroma which contains collagen fibrils and myxoid cells with small stellate, spindle, and round nucleus with long process suggestive of odontogenic fibromyxoma (Figure 3). The patient was prepared and taken up for surgery under local anesthesia. Mass was found



Figure 1: Intraoral view of the myxoma



Figure 2: Cone beam computed tomography (axial section) of the left maxilla

pedunculated from the overlying right alveolar process; local excision was done in Toto. Using a large diamond bur, the margins were smoothened to prevent recurrence as the bone adjacent to the alveolar process was found eroded. The patient was kept on antibiotics and was recalled after 1 month and 4 months (Figure 4).

DISCUSSION

Clinical presentation

Odontogenic fibromyxoma as any other odontogenic tumors occurs in the tooth-bearing areas of mandible and maxilla. Involvement of the mandible is much more common than the maxilla; only a very few cases of fibromyxoma involving the maxilla were reported.^[5]

The tumor arises from the embryonic mesenchymal tissue and is benign in nature; however, when it arises in maxilla, the tumor behaves aggressively, with local bone erosion and sinus invasion causing destruction of maxilla, orbit, and the anterior skull base causing pain and facial deformity.^[6] They can occur anywhere in the jaws but have a predilection for

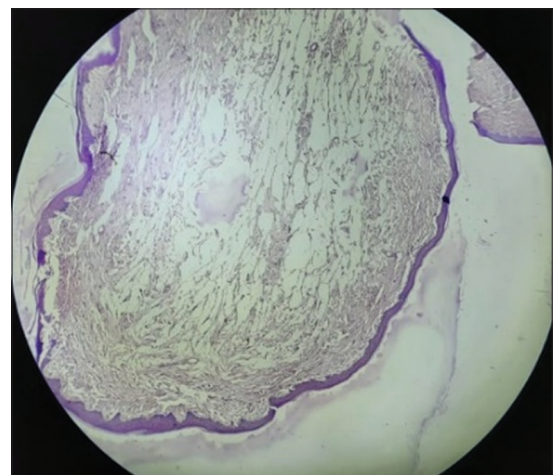


Figure 3: Histopathological view showing collagen fibrils and myxoid cells with small stellate nucleus



Figure 4: Post-operative view after 4 months

the molar and premolar regions.^[7] Patients in the second to fourth decade of their life are affected the most with a little more female predilection.^[8]

In addition, fibromas are mostly asymptomatic, they do not have any malignant potential, and their recurrence is rare after a total excision.

Differential diagnosis: clinical

On the basis of its clinical presentation, the differential diagnosis for fibromyxoma includes a variety of gnathic odontogenic lesions, including ameloblastoma, central odontogenic fibroma, odontogenic keratocysts, vascular conditions (central hemangioma), and non-neoplastic lesions such as central giant cell granuloma. Malignant neoplasms that arise in bone may also be considered.

Imaging characteristics of fibromyxoma

The radiographic presentation of fibromyxoma is variable as they present with multilocular radiolucency with well-defined borders, however, it is common for these lesions to be unilocular.^[9] The multilocular radiolucency has different appearances including “soap bubble”, “honey comb”, “tennis racquet”, “wispy”, or “spiderweb” patterns.^[10] While most odontogenic fibromyxomas present as radiolucency, a small percentage is reported to be radiopaque or mixed radiolucent and radiopaque.^[9]

In our case, the lesion appeared as a large radiolucent area with no trabeculations.

Differential diagnosis: radiographic

Other lesions that appear radiolucent or mixed radiolucent/radiopaque should be considered, including central giant cell granuloma, ameloblastic fibroma, and calcifying epithelial odontogenic tumor. Since approximately one-third of fibromyxomas have poorly defined borders, malignant lesions should also be included in the radiographic differential diagnosis.^[9]

Gross and histopathologic characteristics of fibromyxoma

Grossly, odontogenic fibromyxomas are unencapsulated, well-delineated masses that may be rubbery and soft gelatinous in texture.^[11]

While histologic criteria to distinguish myxoma from fibromyxoma are lacking, microscopic examination of these lesions reveals. A tumor composed of randomly oriented, morphologically bland stellate-to-spindle shaped cells embedded in a loose myxoid extracellular matrix with cells presenting with thin, long cytoplasmic prolongations that give the tissue characteristics of immature mesenchyme.^[12]

The fibro myxoid lesion may present loci of calcification or ossification and a higher amount of collagen fibers and vessels than a typical myxoma.

Differential diagnosis: histopathology

Fibromyxomas should also be differentiated from chondromyxoid fibroma, odontogenic fibroma, desmoplastic fibroma, and myxoid neurofibroma.

Management and prognosis

While there are no universally accepted treatment recommendations for the management of fibromyxoma, it is important to consider the aggressive nature of the neoplasm and make necessary decisions. Although the recommended treatment is usually surgical excision, approaches differ depending on the size of tumor and include conservative surgical excision, enucleation and curettage, curettage with and without electrical or chemical cautery, *en bloc* resection, and wide resection with and without bone grafting.^[13]

Recurrence rates between 25% and as high as 43% are reported for gnathic fibromyxomas.

In some studies, a high rate of recurrence is documented after conservative therapy. This is attributed to the incomplete removal of the lesion rather than the intrinsic biological behavior of the lesion. Due to the tumor’s local aggressive biological behavior and high potential for recurrence, wide surgical resection is recommended. Radiotherapy is generally agreed to be ineffective in the treatment of fibromyxoma. Resection with 1.0–1.5 cm bony margins and one uninvolved anatomic barrier margin is suggested. Follow-up of the patient is warranted due to the high recurrence potential of this tumor.

CONCLUSION

Odontogenic fibromyxoma is a rare, aggressive neoplasm that occurs most commonly in the posterior mandible of females. Histopathologic examination is essential to distinguish this lesion from other benign processes and malignancies to ensure proper patient management.

The radiological examination by means of computed tomography and magnetic resource imaging plays an important role in the diagnosis of a fibromyxoma and in the differential diagnosis from other pathological entities such as ameloblastoma.

The odontogenic fibromyxoma, though not life threatening in the immediate time span, is annoyingly symptomatic and affects the quality of life badly because of its location and nature. Hence, complete removal of the tumor including the involved tooth, adjacent soft tissue, and bone is advised.

Even after a satisfactory surgical resection, prolonged follow-up is advised in view of the high tendency of the tumor to recur.

Consent

The patient’s written and informed consent has been collected and preserved by the author(s).

CONFLICT OF INTERESTS

The authors do not have any relevant conflicts of interest.

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