A rare case report of myxoid fibroma of maxilla

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Abstract: This case report describes about a swelling in maxillary right posterior teeth region. On examination the swelling was solid, pinkish, red, non-ulcerated & non-tender overgrowth at the right maxillary posterior teeth region with no buccal or palatal expansion. Panoramic radiograph revealed a faint radiolucent area with few radiopaque foci and histopathological examination showed myxoid fibroma. Further, correlation of clinical, radiological & histopathological feature; essential to diagnose lesion which lack the characteristic features are also discussed.

Keywords: Myxoid fibroma; odontogenic myxoma; tennis racquet appearance

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Introduction

This is a unique case of myxoid fibroma of right maxilla which brings together the fields of Oral Medicine, Oral Surgery and Oral Pathology.

Odontogenic myxoma is a rare intra-osseous neoplasm, which is benign but locally aggressive. It rarely appears in any bone other than jaws. Soft tissue myxomas are frequently found but their intra-osseous counterparts are rarely encountered and those that are found are almost exclusively confined to the jaws. Odontogenic myxoma most frequently occurs in second or third decades of life. It has a slight female predilection and involves the mandible more commonly than maxilla (1). Clinically, odontogenic myxoma is a benign painless, invasive, slowly enlarging mass causing marked asymmetry of the face. Radiographically, it appears as unilocular pericoronal radiolucency with variable trabecular pattern giving rise to soap bubble, tennis racquet or honey comb appearance. However, other radiological appearances like "Sun-ray" appearance have also been reported in the literature (2).

Thus, the purpose of this article is to report a rare case of myxoid fibroma of right maxilla.

Case presentation

A 52-year-old female reported to the Department of Oral

Medicine with a chief complain of swelling in upper right back teeth region for past 3 years. At the onset, she noticed a swelling measuring about the size of a peanut in the gingiva of upper right back teeth region which gradually increased to its present size. The swelling was asymptomatic and she never noticed any discharge from the effected region. However, she experienced difficulty in mastication as the swelling interfered with the occlusion. She neither consulted any physician nor took any medication for the same and reported to the department for the first time. She had habit of areca-nut chewing 3-4 times a day for past 2-3 years. Past medical & dental history gave no relevant findings and general examination revealed all her vital signs to be within the normal range.

On extraoral examination, a solitary swelling was visible on right side of face which was extending anterioposteriorly from nasio-labial fold to 3 cm away from corner of mouth and superior-inferiorly from canine fossa to corner of mouth. The swelling was round to oval in shape and approx. 2 cm \times 3 cm in dimension with ill-defined borders. Overlying skin appears to be normal with no secondary changes (*Figure 1*). On palpation, overlying temperature was normal. The swelling was non-tender, firm to hard in consistency, non-compressible, non-reducible, nonpulsatile, non-fluctuant and was not fixed to underlying structure. Bilateral submandibular and submental lymph

Quantitative Imaging in Medicine and Surgery, Vol 5, No 5 October 2015



Figure 1 (A,B) Painless bony hard swelling on the right maxilla.



Figure 2 (A,B) Intraoral image showing an overgrowth on maxillary right posterior teeth region extending from distal aspect of 14 to mesial aspect of 17.

nodes were palpable, non-tender and movable.

On intraoral examination, an overgrowth was visible on maxillary right posterior teeth region extending from distal aspect of 14 to mesial aspect of 17 measuring approx. $3 \text{ cm} \times 5 \text{ cm}$ in diameter round to oval in shape with welldefined borders and lobulated surface there by obliterating the right buccal vestibule. Overlying mucosa appeared to be inflamed. On palpation, the overgrowth was nontender, pedunculated, bony hard in consistency, and nonpitting on application of pressure with rolled out margins (*Figure 2*). On the basis of history and clinical findings a



Figure 3 Panoramic radiographs showing a single large expansile radiolucent lesion without any trabeculations in the area of bony destruction, with few radio-opacities seen within the radiolucency.

provisional diagnosis of ossifying fibroma was proposed.

Differential diagnosis: odontogenic myxoma should be included in the differential diagnosis of both radiolucent and mixed lesions of the jaw (3). Honey comb variant of the myxoma usually shows fine trabeculations within the small lobules, which are not present in ameloblastoma (4,5). Giant cell granuloma occurs most commonly in anterior region of the jaw, whereas myxoma is seen most frequently in premolar-molar region (4,5).

Investigations: routine hematological investigations were normal. Panoramic radiograph revealed a single large expansile radiolucent lesion without any trabeculations in the area of bony destruction. However, few radio-opacities were seen within the radiolucency (*Figure 3*).

An incisional biopsy was performed and the specimen on gross examination showed grayish white, glistening smooth, gelatinous mass. Histopathology of the tumor showed typical features of odontogenic myxoma containing thin stretched keratinized stratified squamous epithelium. Connective tissue stroma was fibromyxoid with numerous ovals to plump, spindle shaped cell. At places fibro-stroma was more concentric to lamellar like calcification which was suggestive of myxoid fibroma (*Figure 4*).

Treatment: surgical excision (*Figure 5*) and curettage of the tumor was performed under general anesthesia followed by extraction of maxillary right first premolar. Primary closure was achieved.

Outcome and follow up: healing was uneventful and follow-up after 4 months showed no recurrence of the lesion (*Figure 6*).

Discussion

Virchow in 1863 coined the term myxoma for a group of tumors that had histological resemblance to the mucinous substance

Khan et al. Myxoid fibroma of maxilla



Figure 4 (A) Histological image ($4\times$ view) showing stretched epithelial lining; (B) $10\times$ view showing chondroid like cell in connective tissue stroma; (C) $40\times$ view showing the typical feature of myxoid fibroma having spindle shape cell in myxomatous stroma.



Figure 5 Excised tissues.

of the umbilical cord. In 1947, Thoma and Goldman first described myxomas of the jaws. Odontogenic myxoma has been thought to originate from primitive mesenchymal structures of the developing tooth (follicle/papilla/periodontal ligament) as an inductive effect of nests of odontogenic epithelium on mesenchymal tissue or as a direct myxomatous change in fibrous tissue, hence called odontogenic myxoma (6,7). Myxomas of the head and neck can be identified in two forms (I) facial bone derived, which had been sub divided in the past into true osteogenic myxoma and odontogenic myxoma and (II) soft tissue myxoma derived from perioral soft tissue, parotid gland, ear and larvnx (8,9). Incidence rate is approximately 0.07 new cases per million people per year and are more common in females (10-12). The prevalence of odontogenic myxoma is between 0.04% and 3.7%. In Asia, Europe, and America, relative frequencies between 0.5% and 17.7% have been reported (11). Odonogenic myxoma is a non-encapsulated benign tumor of the jaw bones, comprising around 3-6% of all odontogenic tumors. Previous studies by White et al. (4), reported the incidence with an age range of 11-70 years,



Figure 6 Post-treatment follow up after 4 months.

but the majority of the cases are between 10 and 40 years of age, according to Günhan *et al.* (13). However the present case was reported at the age of 52 years.

The mandible appears to be more frequently affected than the maxilla, especially the posterior region. According to Reichart and Philipsen, mandibular myxomas accounted for 66.4%, with 33.6% in the maxilla. Whereas 73.8% and 65.1% of the cases were located in the molar and premolar areas in maxilla and mandible respectively (14). However, in the present case, the lesion was cited in the premolar and molar area of the maxilla. Odontogenic myxoma of the maxilla behaves more aggressively than that of the mandible while it spreads through the maxillary sinus as presented in our case. Kaffe et al. found expansion of the jaws in 74% of the cases. When the maxillary sinus is involved, the odontogenic myxoma often fills the entire antrum. In severe cases, nasal obstruction or exophthalmus may be the leading symptoms. Displacement of teeth has been registered in 9.5% of the cases (15).

On gross examination of the specimen, the gelatinous,

Quantitative Imaging in Medicine and Surgery, Vol 5, No 5 October 2015

loose structure of the myxoma was obvious. Microscopically, the myxoma is made up of loosely arranged spindleshaped and stellate cells, many of which have long fibrillar processes that tend to intermesh. The loose tissue is not highly cellular, and these cells do not show evidence of significant activity (pleomorphism, prominent nucleoli or mitotic figures). The intercellular substance is mucoid. The tumor is usually interspersed with a variable number of tiny capillaries and occasionally strands of collagen (16-18).

The tumor is not radiosensitive, and surgery is the treatment of choice. Treatment of Odontogenic myxoma varies from local excision, curettage, or enucleation to radical resection depending on tumor size (19-21). The aggressive nature of odontogenic myxoma is well documented in the literature. While generally considered a slow growing neoplasm, odontogenic myxoma may be infiltrative and aggressive, with high recurrence rates. Because of poor follow-up and lack of reports, a precise analysis of recurrence rates is still missing. In the present case, the tumor was completely removed by surgical excision and no recurrence was reported even after 4 months of the surgery.

Conclusions

There is a wide variety in clinical and radiological appearance of odontogenic myxomas, and the most common form of presentation being an asymptomatic expansion in the jaw and a multilocular radiolucent image. Hence correlation of clinical, radiological and histopathological features is essential when trying to diagnose lesions which lack the characteristic appearance. Early detection and complete surgical excision of these lesions followed by long term follow up bears importance in clinical management due to aggressive nature and high recurrence rate. The presented case showed no clinical or radiological evidence of recurrence after 4 months of post excision follow-up. Since the time elapsed from surgery is still short, continued clinical and radiological monitoring is required.

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None.

Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

Consents: We have obtained patients' consent.

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Khan et al. Myxoid fibroma of maxilla

782

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