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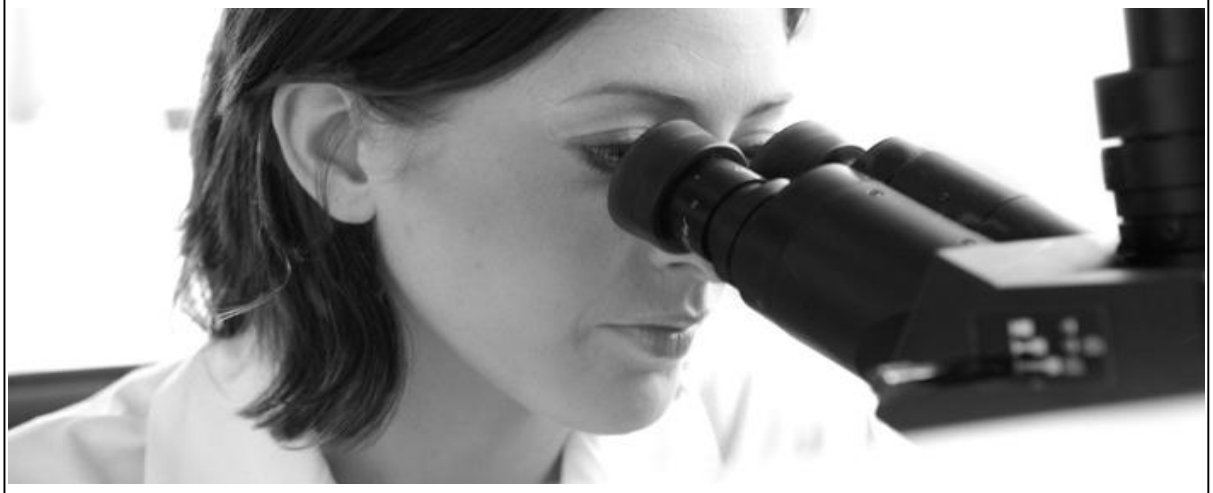

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CASE REPORT

MAXILLARY POSTERIOR ODONTOGENIC MYXOMA -- UNUSUAL OCCURENCE : A CASE REPORT

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ABSTRACT

Odontogenic Myxoma is a benign odontogenic tumor of odontogenic ectomesenchyme with or without included odontogenic epithelium. It is a tumor of the jaw which apparently arises from the mesenchymal portion of the tooth germ either the follicle, dental papilla or the periodontal ligament. It is a central lesion of the jaws which expands the bone and causes destruction. Odontogenic myxoma, an uncommon benign neoplasm comprises about 3–6% of all odontogenic tumors. Some investigators made a distinction between odontogenic myxomas derived from odontogenic mesenchyme and osteogenic myxoma derived from primitive bone tissue. However it has been considered all myxomas of the jaws to be of odontogenic origin.In this article we present a rare case of Odontogenic Myxoma which occurred in the posterior maxilla of a 30 year old female.

INTRODUCTION

Myxomas are considered to be relatively rare benign tumors of mesenchymal origin. Myxomas can be found in the body including the skin and subcutaneous tissue, heart (mainly in the left atrium), and also in various sites of the head and neck.[1] The relative prevalence of odontogenic myxoma (OM) as reported in a recent report was 0.2-17.7% of all the odontogenic tumours and it was made a separate entity in the WHO's classification for histological typing of odontogenic tumours. It is defined as: "a benign tumor, which is ectomesenchymal in origin and is a locally invasive neoplasm consisting of rounded and angular cells lying in an abundant mucoid stroma[2]." Odontogenic myxoma of the jaw, a rare benign tumor is characterized by mucoid or gelatinous grayish-white gross tissue which replaces the cancellous bone leading to expansion of cortex[3]. It is either mesenchymal or ectomesenchymal in origin.[4] OM's are locally invasive non metastasizing neoplasms of the jaws, almost exclusively occuring in tooth-bearing areas and most frequently seen in the second or third decades of life.[5,6] Children and elderly persons are seldom affected.

Myxomas can occur anywhere in the jaws but have a predilection for the molar and premolar regions of the mandible. Cortical expansion and perforation are common findings and maxillary myxomas could even extend into the maxillary sinus.[7] Jaw bone myxomas are traditionally considered to be odontogenic in origin. Past reported literature states that OM's represent between 1% and 17.7% of all odontogenic tumors. Microscopically these lesions are characterized by stellate shaped cells embedded in a richly myxoid extracellular matrix, with little collagen; those cases with higher amounts of collagen may be denominated as myxofibroma. These neoplasms are said to be derived from the mesenchyme of a developing tooth or from the periodontal ligament. Islands of inactive odontogenic epithelium may be found in a few cases, but there is no proof of them exerting inductive effects over the surrounding mesenchyme. The exact nature of OM is unknown, but few studies state that the cells and extracellular matrix of OM are different from the ectomesenchymal tissues of developing tooth.[8] Radio graphically, the tumor appears as well-defined unilocular or multilocular radiolucency with fine bony trabeculae within its

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interior structure giving it a `honeycombed', `soap bubble', or 'tennis racket' appearance. Unilocular OM's are seen more in the anterior part of jaws and in children. The tumor often scalloped between the roots causes teeth displacement commonly and root resorption is rarely seen.[9] Our article highlights a rare case of odontogenic myxoma of the maxilla in a female and its varied histopathological features have also been discussed.

Case Report

A 30-year old female reported to the OPD of our college with a complaint of a painless swelling in the posterior region of maxilla for the past 6 months. Initially the swelling and growth was small in size which gradually increased to its present size. The swelling and growth extended from first molar region to the maxillary tuberosity posteriorly, and it obliterated the buccal vestibule (Fig 1).

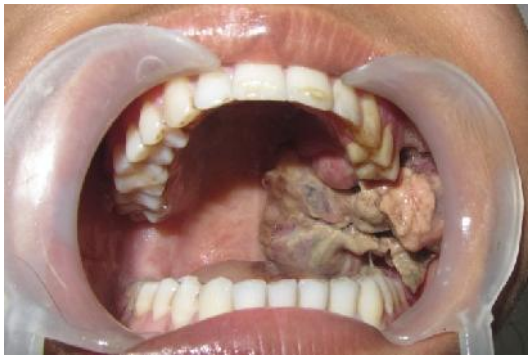

Fig 1 Intraoral picture, growth extending from first molar anteriorly to tuberosity region posteriorly.

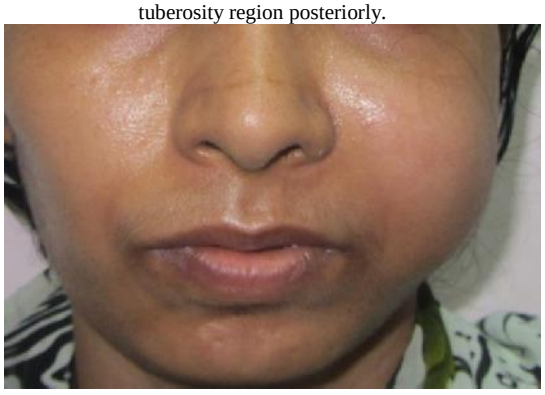

Fig 2 Extraoral picture, bony hard, tender swelling in upper left side of face.

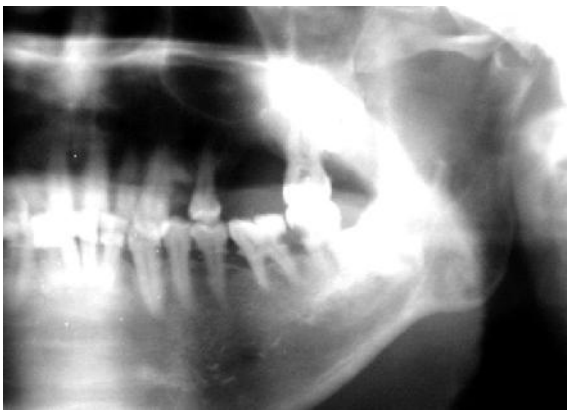

Fig 3Unilocular radiolucency with irregular margins involving maxillary antrum.

A biopsy was performed along with which two associated teeth were extracted and the result was not known. Extraoral examination showed diffuse swelling in the left infraorbital region, obliterating the naso-labial fold (Fig 2). The skin over the swelling and local temperature was normal. The swelling was hard anteriorly and felt soft in the posterior region on palpation. Buccal and palatal cortical expansion was seen and there was no history of paresthesia. Orthopantomograph showed a well-demarcated unilocular radiolucent lesion with irregular margins involving the maxillary antrum(Fig 3). Histopathological examination of incised biopsy specimen revealed dense and diffuse collection of collagen fibers and fibroblasts and small inconspicuous strands of odontogenic epithelium.

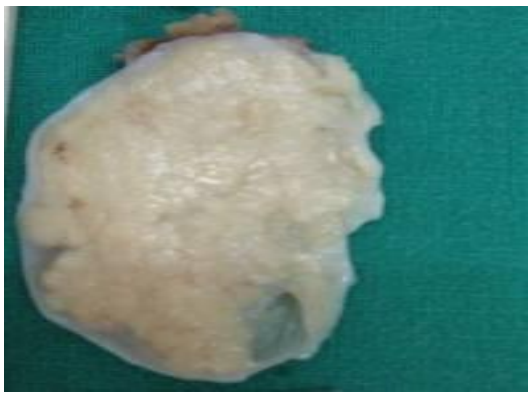

Fig 4 Gross excised specimen showing white gelatinous texture.

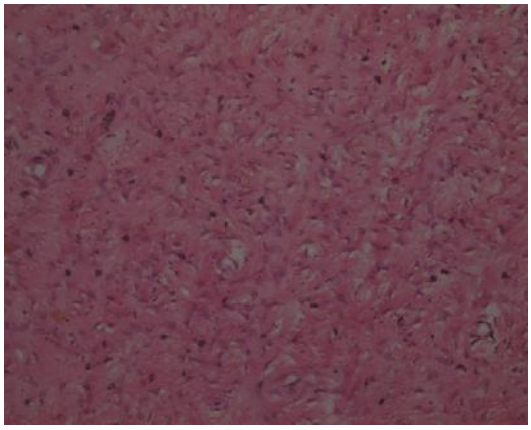

Fig 5 Micrograph shows collection of fibroblast with inconspicuous strands of odontogenic epithelium in a myxoid stroma; (H/E; 10X)

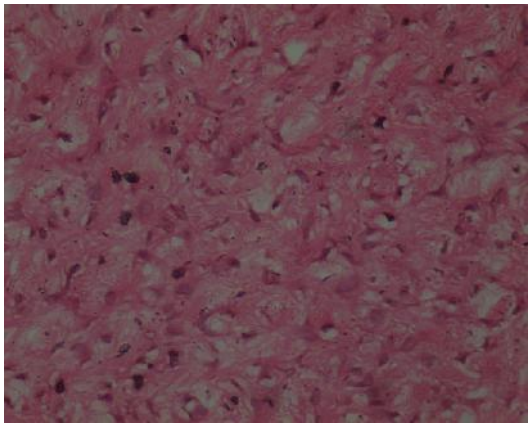

Fig 6 Micrograph shows mixed area of fibrous tissue and strands of odontogenic epithelium in a myxoid stroma; (H/E; 20 X)

It was diagnosed as odontogenic fibroma, and the lesion was surgically excised. Gross tissue examination revealed the lesion to be white in color with a soft gelatinous texture (Fig 4). Histopathologically the tissue revealed an unencapsulated tumor mass with spindle and stellate shaped cells in a loose myxoid connective tissue stroma, which beared close resemblance to the mesenchymal portion of a developing tooth. Few areas showed moderately dense collagen fibers and strands of odontogenic epithelium were seen in the tissue stroma. The overall impression of the lesion was more amount of myxoid stroma in less fibrous and acellular background (Fig 5, 6). This histological feature of excised specimen was different from that of the incisional biopsy, which was highly cellular and fibrous in nature, that had led to the diagnosis of odontogenic fibroma. The presence of myxomatous tissue along with areas of moderately dense collagen fibers, led to the final diagnosis of Odontogenic Myxoma.

DISCUSSION

Odontogenic myxoma (fibromyxoma), a benign odontogenic neoplasm with uncertain histogenesis has a characteristic histologic appearance. It is locally aggressive and shows infiltration.[7] Though it was originally described by Thoma and Goldman in the year1947[10] but the nature of OM has been ambiguous till date. Many scientists have performed several studies to define its precise nature, but at present there is no universally accepted theory about its probable histogenesis. OM is odontogenic in origin chiefly from dental follicle or from periodontal ligament has been supported by the presence of occasional small islands of odontogenic epithelium, its occurrence almost exclusively in the jawbones and the histomorphological similarity to the mesenchymal component of the developing tooth.[11,12]

Biochemical composition of OM is said to be totally different as compared to the mesenchymal dental tissues, raising doubts whether to regard it of true odontogenic origin. In this respect Slootweg *et al*[13] found significant differences on specific glycosaminoglycan (GAG's) content, such as that hyaluronic acid in OM was four times higher than other GAG's, such as chondroitin sulfate, which was inverse to what has been observed in dental pulp, gingival tissue and periodontal ligament. This fact was supported by the findings of Sakamoto et al and Embery who studied the GAG's contents found that major GAG's in OM were chondroitin 4 and 6 sulphates whereas in human dental pulps it was dermatan sulfate which was present in higher proportion. The cellular population in OM is heterogeneous, but several studies do state that this neoplasm is mainly composed by actively secreting fibroblastic cells, as well as a significant number of myofibroblasts (MF). "Myxoblast" was a term once used to name the modified fibroblasts but it seems that these cells belong to the myofibroblastic category.[8]

Head and neck myxomas are rare tumors which have two forms: (1) facial bone derived, further divided into true osteogenic myxoma and odontogenic myxoma and (2) soft tissue derived myxoma, derived from the perioral soft tissue, parotid gland, ear and larynx.[14] Myxomas of the maxilla and mandible are considered to be neoplasms of odontogenic origin supported by the facts that they almost exclusively occur in the tooth-bearing areas of the jaws, their common association with an unerupted tooth or developmentally absent tooth, their frequent occurrence in younger individuals, their histologic resemblance to dental mesenchyme, especially dental papilla and the occasionally seen parts of odontogenic epithelium.[15] Slootweg and Wittkampf in their study showed that the dental pulp and PDL matrix is completely different from the jaw myxomas matrix. They also stated that myxomas could have a non-odontogenic origin too as they also develop in the sinonasal tract and other facial bones. They were of the opinion that the presence of odontogenic epithelium was not even necessary to make the diagnosis of myxoma of bone. [16]

In a review of over 600 bone tumors of patients at Mayo Clinic it was concluded that no true myxomas of the bone existed except those found in the mandible and maxilla; they were contrary to the case report findings of Slootweg and Wittkampf.[17]

A study conducted on 164 odontogenic myxomas of the jaw revealed that 75% occurred between the second and fourth decades (patient age range, years).[18] Farmanÿ et al differentiated maxillary and mandibular OM's and stated the mean age of maxillary odontogenic myxomas in men was 29.2 years and in women was 35.3 years at the time of diagnosis, while the mandibular odontogenic myxomas in men occur at a mean 25.8 years and in women they occur at 29.3 years[6]. A higher incidence of OM's was reported in women (64–95%) than in men[6]. Jaw myxomas occur 66.4% in the mandible (65.1% located in premolar and molar areas) and 33.6% in the maxilla (73.8% cases were seen in the same areas of maxilla).[19] Our case reported the lesion was located in the premolar and molar area of the maxilla.

Most OM's are usually first noticed as a slowly increasing swelling or jaw asymmetry. They are usually painless in nature and seldom ulcerate when they interfere with dental occlusion. Growth may be rapid and infiltration of neighbouring soft tissue structures may occur. Buccal and lingual cortical plates of the mandible may show occasional expansion.[20]

Another study found jaw expansion in 74% of the reported cases. Of the cases infiltrating the maxillary sinus, OM's filled the entire antrum. Nasal obstruction or exopthalmus were leading symptoms of OM's of the antrum. Although exopthalmus was not noticed in the present case, the right lower eyelid and the eyeball were pushed upward. Teeth displacement was noticed in 9.5% of the cases. Jaw myxoma radioluscencies are usually multilocular either "honey comb," "soap bubble" or "tennis racket" in appearance and they are differentiated from malignant tumors which arise centrally from the jaw bones, because the latter usually cause massive bone destruction without compartments formed by bony trabeculations or bony septa.[21] In our case, OPG revealed a single, large expanding radiolucent lesion with no bony trabeculations in the area of destruction, though few radio opacties were seen within the radiolucency. Gross tissue examination of the specimen revealed the gelatinous, loose structure of the myxoma. Microscopically, the myxoma was made of loosely arranged spindle-shaped and stellate cells,

which had intermeshed long fibrillar processes. The loose tissue was not highly cellular, and cells showed no evidence of significant activity. The intercellular substance was mucoid. The tumor was interspersed with tiny capillaries and occasionally strands of collagen. The amount of collagen fibrils are more prominent in fibromyxoma and they can be best demonstrated by silver impregnation method identifying them as reticulin fibres. Odontogenic epithelium remnants are infrequently noted, sometimes being surrounded by a narrow zone of hyalinization. The myxomatous component of OM's resembles the primitive mesenchyme, dental papilla and dental follicle.[6] Two basic cell types of tumor cells namely secretory and nonsecretory have been described in detail in an extensive study done on the ultrastructure of odontogenic myxomas in 1976. The secretory cell type was the principal tumor cell and resembled fibroblasts.[19]

OM's are slow-growing in nature and hardly show infiltration or recurrence. Precise data of recurrence rates is still missing due to poor follow-up and lack of reports. OM's could be treated by local excision, curettage, enucleation or radical resection. Recurrence if present could be due to the type of therapy, with conservative surgery resulting in a higher number of recurrences. In our case, the tumor was removed completely by surgical excision and post 5 months of the procedure no recurrence has been reported.

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