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Odontogenic Myxofibroma of the Jaw: A Case Report

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ABSTRACT

Odontogenic Myxofibroma is one of the comparatively rare benign mesenchymal odontogenic tumors in the jaw with a high recurrence rate. A 28-year-old female patient reported frequent pain for over one year. Extra-oral examination and Intra-oral examination were insignificant. Orthopantomography (OPG) showed a radiolucent lesion in the left canine and premolar region. Computed tomography scanning reveals a well-defined reveal radiolucent multilocular cystic cavitary lesion with an intact outer border of the mandible, and resorb of the lingual cortex. Multiple small irregular pieces of this lesion were enucleated (excisional biopsy) and sent to the Pathology department for examination. The histopathological assessment revealed loose fibromxyomatous tumors exhibit a proliferation of spindle-shaped cells that produced wide mucoid-rich intercellular stroma(myxoid areas), consistent with Odontogenic Myxofibroma. Immunohistochemistry plays a great role in confirming and ruling out other differential diagnoses. The choice of treatment depends on certain variables such as localization, presence of a primary or recurrent lesion, age, general medical conditions, and aesthetic needs of the patient. The best management strategy applied in this patient is total enucleation of the lesion along with curettage of the cavity.

Key words: Odontogenic tumors, Myxofibroma, Myxoid.

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INTRODUCTION

Odontogenic Myxofibroma\Myxoma is a comparatively rare entity of benign, painless, slow-growing mesenchymal (intraosseous) spindle cell tumor, considered as a variant of odontogenic myxoma. It's included in the latest updated publication of the WHO classification of head and neck tumors (2017), as a separate category under the Mesenchyme and/or odontogenic ectomesenchyme with or without odontogenic epithelium (4th edition) [1]. These tumors are considered the third most common odontogenic tumor, most frequently encountered in females between the second and fourth decades, located mainly in the mandible, particularly in the posterior region with a high rate of recurrence [2]. Only a few studies calculate the incidence rate of this condition, however many studies described the clinical course and clinical presentation of the Myxofibroma or Fibromyxoma or Myxoma [3].

Generally, Odontogenic Myxofibroma consists of proliferative bland-looking spindle cells with focal intervening accumulation of abundant myxoid or mucoid intercellular matrix [4].

Analysis performed on a few soft tissue and intraosseous tumors, with focal loose myxomatous changes found to be rich in acid muco polysaccharides\glycosaminoglycan, hyaluronic acid, and in many body sites, this is considered a degenerative phenomenon but it also contains considerable amounts of collagenase fibrous stroma deposition [5].

Fibromyxoma/Myxofibroma, Odontogenic fibroma, and myxoma are considered

histogenetically as one heterogenous spectrum of benign mesenchymal tumors, but behaviorally are distinct [6].

We present a case of Myxofibroma, which has a unique and unusual presentation in a 28-year-old temperature rise or change in the overlying skin color female at the left side of the jaw.

CASE REPORT

A 28-year-old female patient reported to the maxillofacial clinic with a history of frequent pain on the left side of the lower jaw, for the past year. There was no history of trauma and the past dental& medical history was noncontributory. Extra-oral examination reveals no evidence of apparent swelling detected. No systemic or local rise in temperature or change in color of the overlying skin was noted. No lymphadenopathy was observed. No paresthesia of the inferior alveolar nerve was detected. No swelling, erythema, ulceration, or pus discharge was detected on intraoral examination.

Radiographically, Orthopantomography (OPG) showed multilocular radiolucent lesions in the left canine and premolar region, with no relation to the apex of the teeth [Figure 1].

CT scan reveals a well-defined radiolucent cystic cavitary lesion with an intact outer border of the mandible and resorb of the lingual cortex [Figure 2 A-D].

The patient was prepared to the Operating Room (OR) and operated under general anesthesia for enucleation of the cystic lesion. The operation started by giving infiltration local anesthesia in the labial and buccal mucosa. A surgical incision was performed extending from mid-line to the premolar region in the left sulcus. Then the mandible bone was exposed, and the buccal cortical bone was found intact. The lesion was enucleated easily with good exposure to the area using a round bur [Figures 3, 4] the lingual cortex was found resorbed and the contact with the lingual mucosa. Excisional tissue biopsy was sent to histopathology with provisional diagnoses of odontogenic myxoma, myxoid neurofibroma, ontogenetic fibroma, desmoplastic fibroma, and chondromyxoid fibroma.

The gross pathology of the surgical specimen received consists of irregular small pieces of soft tissue, brownish to tan-white in color, measuring 1 cm in aggregate diameter. Histologically, the tumor was composed of proliferative mixed fibro-collagenous and myxoid stromal oval-spindle cells with alternate hypo cellular and hyper cellular areas. The tumor exhibits the proliferation of bland-looking mesenchymal spindle-shaped cells that produced wide areas of mucoid-rich intercellular collagenase and loose myxoid stroma showing bland spindle cells organized in fascicles oriented in different directions. The nuclei are spindle to oval and some are wavy in shape. The mitotic figure is less than 1 per 10 HPT scrutinized in most cellular areas. No definite odontogenic epithelium rest, dystrophic calcification or significant inflammatory cells were detected. No specific infection or granulomatous reaction was noted. No evidence of cytological atypia or malignancy was seen [Figures 5-10]. Based on the histo morphological architecture, the final diagnosis was given as odontogenic Myxofibroma.

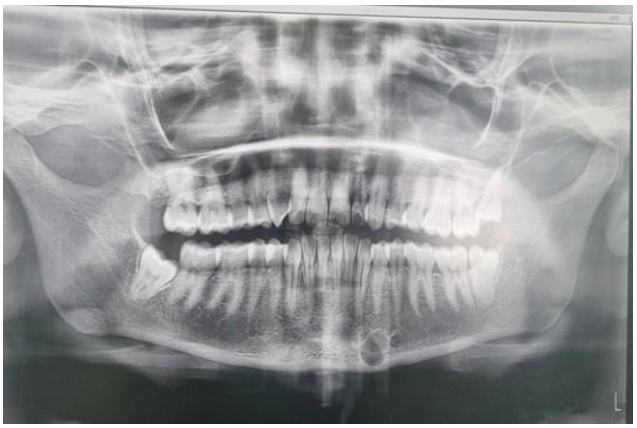

Figure 1: Orthopantomography (OPG) showed a multilocular radiolucent lesion, with a well-defined sclerotic margin in the left canine and premolar region, with no relation to the apex of the teeth.

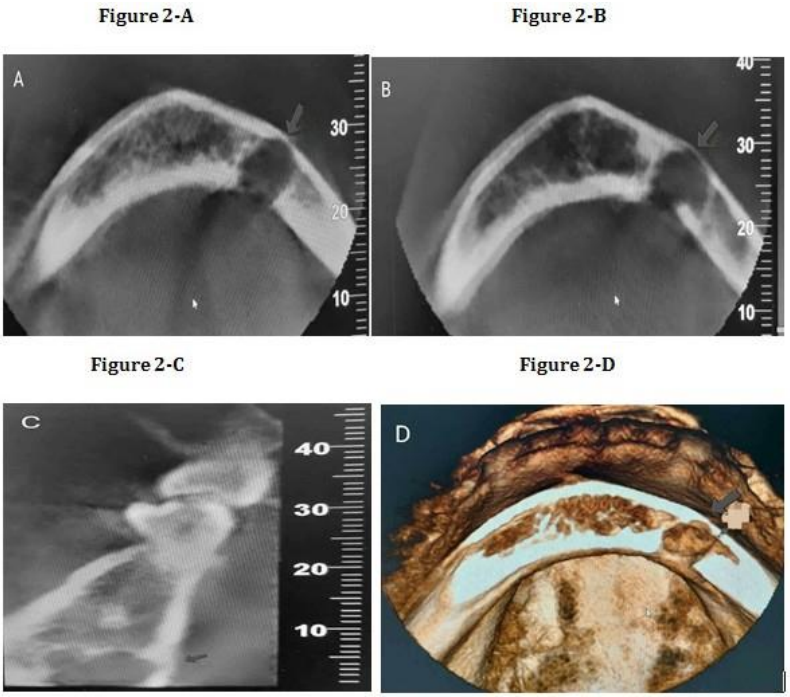

Figure 2: CT scan (A, B: axial view, C: sagittal view and D: 3D reformat) reveals radiolucent cystic cavity lesion in the left canine and premolar region with an intact outer border of the mandible and resorb of the lingual cortex (black arrows).

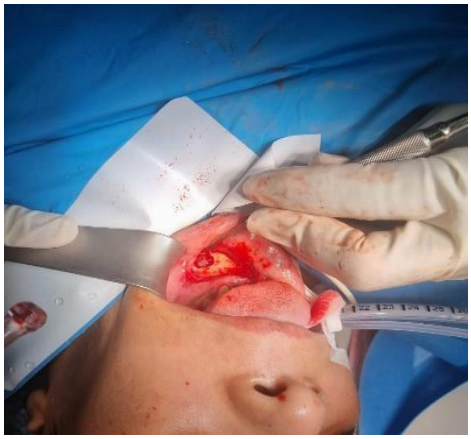

Figure 3: Intraoperative image showing the lesion before removal.

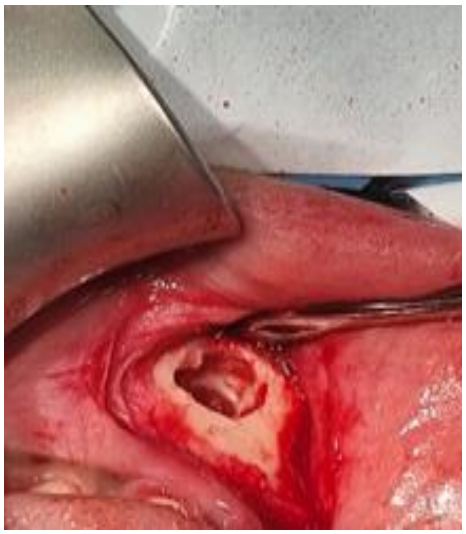

Figure 4: The Intraoperative clinical picture shows the enucleation and removal of the lesion.

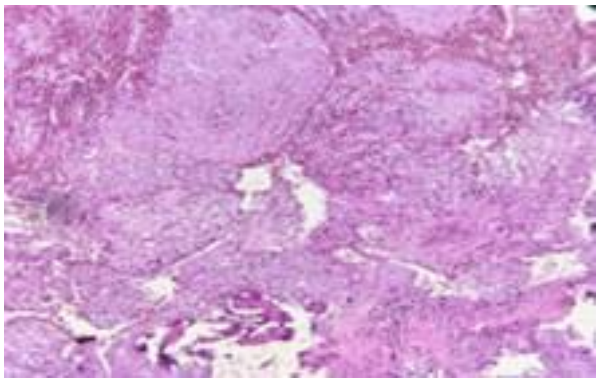

Figure 5: Histopathology section (low power view) showing a lobulated tumor with multinodular growth of proliferative fibrous/fibroblastic tumor, displaying bland spindle cells, arranged in a vaguely whorled growth pattern [H&E section].

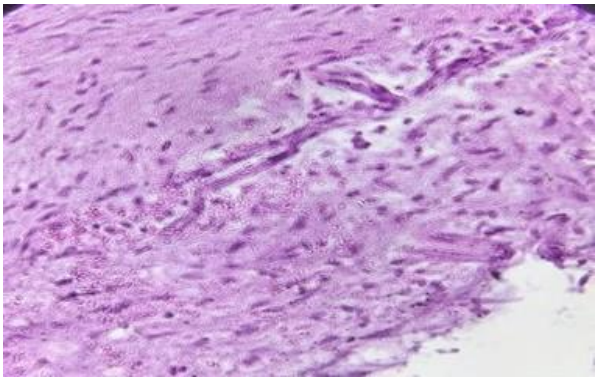

Figure 6: Higher power showed focal areas show cellular wavy thin spindle fibers with no obvious pleomorphic. Mitotic activity is less than 1 per 10 HPF. No odontogenic epithelial islands [H&E section].

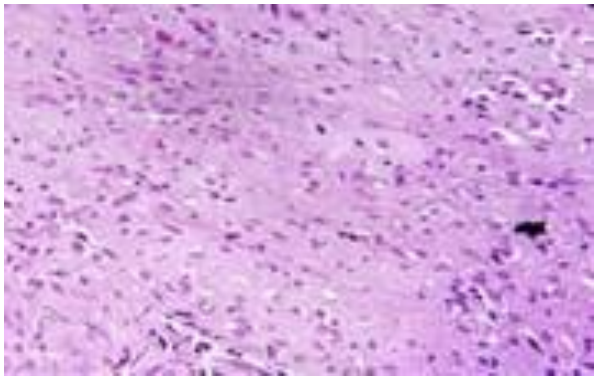

Figure 7: Histopathology microscopic picture reveals sheets of proliferative bland-looking mesenchymal oval-spindle cells; that demonstrate alternate hypo cellular and hyper cellular areas [H&E section].

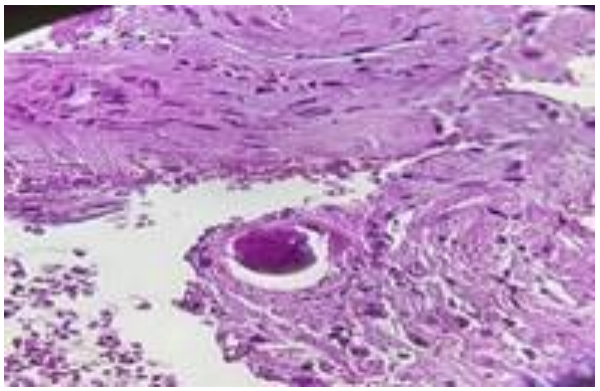

Figure 8: In focal areas, the proliferative spindle fibers start to produce organized collagen clumps. No cellular atypia or mitotic figures are observed [H&E section].

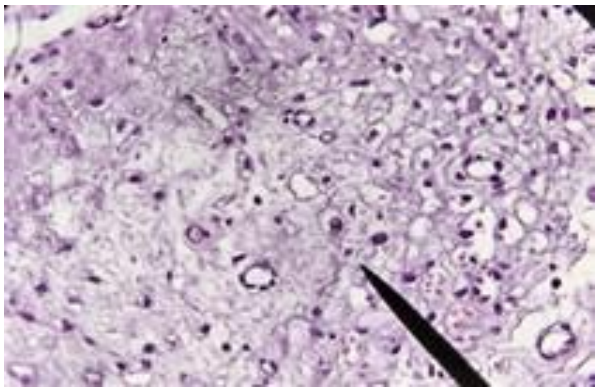

Figure 9: Wide areas of abundant myxoid and edematous stroma with obvious vacuolated fibroblasts [H&E section].

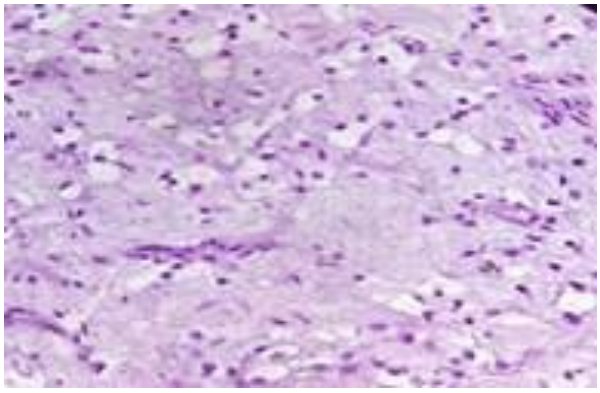

Figure 10: Hypo cellular non-cohesive, bland-looking spindle to oval cells within the myxoid stroma [H&E section].

DISCUSSION

The Odontogenic Myxofibroma (OM) of the jaw is a rare benign mesenchymal soft tissue tumor, characterized by slow-grading proliferative spindle cells with focal intervening myxoid areas. The pathogenesis is still unclear and unknown. There is a lot of debate about the histogenesis of this tumor; some authors considered Myxofibroma to be originated from the mesenchyme of the developing tooth or periodontal ligament and other groups of authors claimed that it's composed of secretory and non-secretory cells that manifest in the intercellular and extracellular deposition of this mucoid/myxoid substance with the presence or absent of odontogenic epithelium [7]. The incidence of this tumor is found to be 0.05 new cases per million population per year, 4 however many studies failed to calculate the incidence rate of Myxofibroma [8].

Clinically, Myxofibroma is found at different sites of the face but is frequently encountered in the tooth-bearing part of the jaw. It has a slow-growing pattern with local aggressiveness (non-encapsulated) if exists for a long duration with adjacent bony destruction [9]. Smaller lesions are usually asymptomatic and discovered during routine radiographic examinations, while large lesions are often associated with painless jaw expansion and possible perforation of the cortical plate. Facial deformity as well as the involvement of the maxillary sinus has rarely been reported [10].

Radiographically, Myxofibroma appears as a localized, well-defined, and more often multilocular than unilocular radiolucent lesion with soap-bubble architecture. Occasionally, in the long-standing clinical course of this lesion, adjacent bone destruction might be found. Gross pathology of Myxofibroma, usually received friable pieces of a white-tan color non-encapsulated fibrous lesion, soft in consistency with mucinous gelatinous appearance. The consistency relies on the amount of collagen or fiber element deposition.

Histopathology demonstrates almost uniform sheets of the spindle to wavy bland-looking cells with alternate cellular to acellular patterns. No mitosis or necrosis was identified. Further Cyto chemical stains (Alcian blue and Periodic acid Schiff-PAS) will help to demonstrate the mucous-

rich intercellular and extracellular material. The Immunohistochemistry workup plays a central role in verification and confirmation e.g.Ki-67, NSE, S100, CD34, Desmin, SMA, MSA, EMA, Calretinin & MUC4. In addition, Immunohistochemistry has additional value in sorting out and analyzing the differential diagnoses of this condition.

Proper diagnostic assessment of this type of tumor (Myxofibroma, myxoma, or Fibromyxoma) is very crucial, because of variable degenerative changes encountered in different lesions/ tumors manifesting in obvious wide areas of myxomatous changes [11]. It's important to distinguish between this variant and other differential diagnoses e.g. Myxoid Neurofibroma (MNF), Chondromyxoid fibroma, Odontogenic fibroma, and desmoplastic Fibroma [12].

The choice of treatment should depend on variables such as localization, presence of a primary or recurrent lesion, age, general medical conditions, and aesthetic needs of the patient. The best treatment option for Odontogenic Myxofibroma of the jaw is complete curettage, enucleation, or complete surgical excision. A high recurrence rate estimated between 25% to 43% was reported due to residual remnants of the tumor as an outcome of incomplete excision.

CONCLUSION

In conclusion, Odontogenic Myxofibroma is a rare slow-growing benign soft tissue tumor that has an indolent clinical course with local aggressive behavior. It has a low rate of recurrence if the tumor is excised and enucleated completely with no residual remnants. The final diagnosis of Odontogenic Myxofibroma will be established after histopathological and Immunohistochemical examination, however, the histo morphological appearances are not pathognomonic, further careful clinical and radiological correlation along with the histopathological features are strongly recommended for proper diagnostic evaluation.

Conflicts of Interest

The authors declare no conflicts of interest.

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