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Odontogenic Myxoma: A Case report and Literature review

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I. INTRODUCTION :

Odontogenic myxoma is a rare locally aggressive benign tumor of the jaw. It accounts for 1–17.7% of all odontogenic tumors. It is asymptomatic and slow growing, characterized by stellate and spindle-shaped cells embedded in an abundant myxoid or mucoid extracellular matrix. It is thought to be derived from the mesenchyme of a developing tooth or the periodontal ligament. Histologic similarity to the pulpal ectomesenchyme, proximity to the tooth-bearing areas of the jaws, periodic association with missing or impacted teeth, presence of inactive odontogenic epithelium in a minority of cases suggests its odontogenic origin. The lesions are not encapsulated, allowing substantial infiltration into the adjacent bone. Consequently, odontogenic myxoma is generally managed surgically; however, there has been some debate as to the most appropriate surgical approach. Surgical management of odontogenic myxoma vary from simple enucleation and curettage to segmental resection. It has high recurrence rate of approximately 25% especially when a more conservative approach is taken. Nonetheless, a more conservative approach exemplified by enucleation and curettage has several advantages over more radical approach. There are currently no clear evidence-based surgical management guidelines for odontogenic myxoma. Here, we describe a case of mandibular odontogenic myxoma managed by enucleation and curettage, in the context of a systematic review of the literature.

II. CASE REPORT :

A 62-year-old man was referred to our department of Oral and Maxillofacial Surgery with the chief complaint of a painless left gradually enlarging maxillary swelling that he noticed 4 months before the initial presentation.The patient reported no symptoms like pain or paraesthesia and the oral mucosa appeared normal. However, an IOPA revealed an extensive radiolucent and multi-locular area with imprecise borders exhibiting a “soap bubble” appearance. Computed axial tomography imaging showed an area of infiltration showing mixed radiopaque radiolucent lesion on the left side of maxilla and also involving the maxillary sinus. The tumor measured approximately $30 \times 15 \times 40$ mm. The patient’s medical history was otherwise unremarkable. An incisional biopsy showed loosely arranged spindle-shaped cells in a myxoid fibrous stroma. On the basis of these histological findings, a provisional diagnosis of odontogenic myxoma was made.

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Fig1:Extraoral Photograph

Fig 2: Intraoral photograph

Fig 3: IOPA showing "soap bubble" appearance

Fig.4:Computer tomography showing mixed radiopaque radiolucent lesion on the left side of maxilla involving the maxillary sinus

We performed extraction of tooth #25, and an excision of the tumor and wide curettage of the normal surrounding tissue under general anesthesia with nasopharyngeal intubation.

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Fig 5:Intra-operative Photograph

The surgical specimen revealed benign-looking spindled and stellate cells in the mucinous stroma. Taken together, these findings confirmed the diagnosis of odontogenic myxoma. There have been no clinical or radiological signs of recurrence over 7 years follow-up.

Fig 6: Excised Specimen

Fig 7: Histopathological Slide showing spindled and stellate cells in the mucinous stroma

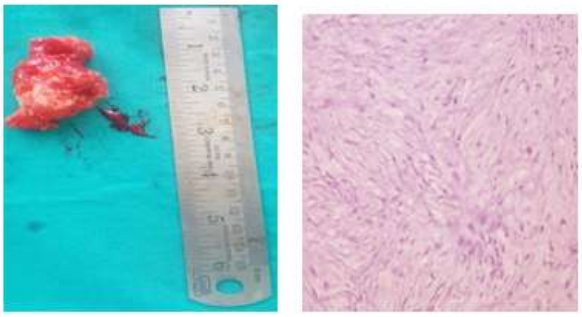

Fig 8:Post-operative photograph

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III. DISCUSSION :

In 1947 Thoma and Goldman first mentioned the term "Odontogenic myxoma" in the literature.It was redefined based on histologic criteria for myxoma by Stout in 1948 as "true neoplasms that do not metastasize and exclude the presence of recognizable cellular components of other mesenchymal tissues, especially chondroblasts, lipoblasts, and rhabdomyoblasts."

In 1992, WHO classified OM for histological typing of odontogenic tumors: "A benign tumor, which is of ectomesenchymal origin with or without the presence of odontogenic epithelium."As an osseous entity, however, OMs of the jaws are considered slow-growing tumors with the potential for extensive bone destruction, cortical expansion, and a relatively high recurrence rate. Odontogenic myxoma is usually a slow-growing mass with late-appearing symptoms, primarily due to the mass effect. Symptoms include pain, paresthesia, ulceration, and tooth mobility (Gonzalez-Garcia et al., 2006), although none of these symptoms were found in the present case.

Although benign, odontogenic myxoma is invasive into surrounding normal bone, sometimes breaking through its boundaries (Chrcanovic et al., 2010). This invasiveness has been attributed to the expression of matrix metalloproteinases 2 and 9, which degrade the extracellular matrix (ECM). These enzymes reportedly cause tumor cells to penetrate the bony trabeculae by acting on the ECM, thus aiding tumor growth (Miyagi et al., 2008, Mauro et al., 2013).

Margins of the lesion are classified as corticated, noncorticated, poorly defined, or diffuse (Noffke et al., 2007). On radiographs the tumor may be uni- or multi-locular (Lo Muzio et al., 1996, Altug et al., 2011), with multiloculated lesions being larger than unilocular ones (Kaffe et al., 1997). The appearance is variably described as mottled, soap-bubble (Zarbo, 2010), tennis racquet (Noffke et al., 2007), or honeycombed (Shafer et al., 2003).In our case, the radiological appearance was of a multilocular, mixed radiolucent-radiopaque type with corticated margins.

Surgery is the treatment of choice, with the treatment protocol depending on the site and size of the tumor. Complete extirpation of the tumor is difficult because infiltration may be more extensive than that observed clinically. Surgery types vary from enucleation and curettage, wide excision, and resection, to radical surgeries involving resection of adjacent tissues (Halfpenny et al., 2000). Allphin et al. (1993) recommended an initially conservative approach, followed by radical surgery if required. In the present case study, an incisional biopsy was performed to confirm the diagnosis and render best possible treatment to the patient followed by wide local excision of the tumor and curettage.

Odontogenic myxoma is radioresistant (Shafer et al., 2003). Although a few researchers advised pre or postoperative radiotherapy (Attie et al., 1966, Cuestas-Carneiro et al., 1988), the present consensus is that radiotherapy has no role in the management of odontogenic myxoma.

Following is the table of review of few earlier studies on odontogenic myxoma:

Researcher	Type of study	Sample size			Range of age in yrs (peak incidence in brackets)	Peak in_decade	Sites		Tooth displacement	Root resorption
		Total	Male	Female			Maxilla	Mandible		
Kaffe et al.	Systematic review with two case reports	164 (96-radiological)	64	100	01–73 (most cases in 2nd to 5th decade)	2nd	55 (33.5%)	109 (66.5%)	26%	9.5%
Martinez-Mata et al.	Retrospective	62	19	43	09–71 (most in 2nd	3rd	25 (40.3%)	37 (59.7%)	12 (19.3%)	Not mentioned

Researcher	Type of study	Sample size			Range of age in yrs (peak incidence in brackets)	Peak in_decade	Sites		Tooth displacement	Root resorption
		Total	Male	Female			Maxilla	Mandible		
					to 4th decade)					
Zhang et al.	Retrospective - radiological	41	22	19	04–63 (most cases in 1st to 5th decades)	3rd	17 (41%)	24 (59%)	21a	10a
Simon et al.	Prospective	33	12	21	03 months–64 years (majority in 2nd to 4th decade)	3rd	08 (25%)	24 (75%)	Number not mentioned	10 out of 21 avlbl cases
Noffke et al.	Retrospective	30	09	21	11–70 (most cases in 2nd to 3rd decade)	3rd	11 (36.7%)	19 (63.3%)	22 (73%)	13 (43%)
Ajike et al.	Retrospective	27	8	19	11–70 (peak in 4th decade)	4th	13 (48%)	14 (52%)	Not mentioned	Not seen
Li et al.	Retrospective	25	13	12	06–66 (peak between 2nd and 5th decade)	3rd	13 (52%)	12 (48%)	11	03
Friedrich et al.	Retrospective - radiological	14	3	11	08–45	–	5 (35.7%)	9 (64.3%)	8	2
Lo	Retros	10	3	7	15–65	4th	4 (40%)	6 (60%)	2 of 10	2 of

Researcher	Type of study	Sample size			Range of age in yrs (peak incidence in brackets)	Peak in_decade	Sites		Tooth displacement	Root resorption
		Total	Male	Female			Maxilla	Mandible		
Muzio et al.	pective									10
Abiose et al.	Retrospective	10	2	8	10–40 (All between 2nd and 5th decade)	3rd	4 (40%)	6 (60%)	Number not mentioned	Number not mentioned

IV. SUMMARY AND CONCLUSIONS:

In respect of biological behavior and extensiveness of Odontogenic Myxoma, better knowledge, correlation of clinico-radiographic appearance with histologic counterpart are mandatory for such lesions to avoid controversies and to reach the final diagnosis and to prevent further recurrences.

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