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Case Report

Peripheral myxoma of the infratemporal region: An unusual case report

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ABSTRACT

Odontogenic myxoma (OM) is a rare locally invasive benign neoplasm, almost exclusively occurring in the jawbones, comprising 3–6% of all the odontogenic tumors. The mandible is more commonly involved than the maxilla. Intraoral soft tissue myxoma is an extremely rare lesion and only few reports are available in the literature. We present probably the first of its kind in literature a soft tissue OM occurring in an unusual location: the infratemporal fossa.

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1. Introduction

Odontogenic myxoma (OM) is a locally invasive odontogenic ectomesenchyme neoplasm.¹ Odontogenic myxomas can be classified into two types, central myxoma located in the bone or peripheral myxoma located extraosseously in the soft-tissue overlying the tooth-bearing areas.^{2,3}

Intraoral soft tissue myxoma is an extremely rare lesion and only a few reports are available in the literature. In this

article, we present the rare occurrence of an extra osseous, soft tissue myxoma in a very unusual location, i.e., the infratemporal fossa region.

2. Case report

An 11-year-old male child reported to our Maxillofacial OPD with the complaint of a swelling on the left side of the face of 1-year duration, which had gradually increased in size (Fig. 1).

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Fig. 1 – (a) Clinical extraoral frontal photograph demonstrating swelling in the left cheek area. (b) Axial CT view demonstrating mass arising from left infratemporal region showing displacement of left maxillary posterior wall and resorption on medial aspect of left ramus.

The patient also gave a history of pain in the left maxillary teeth on mastication. His medical history was noncontributory.

Extra oral examination revealed the face to appear grossly asymmetrical. A well-defined hard swelling, 2 cm × 2 cm × 2 cm, was noted in the left maxillary region. The skin over the swelling was normal. The oral opening was slightly decreased. Intraorally, a firm swelling involving the left maxillary region, as well as some portion of the posterior pterygopalatine region, was noted. The swelling was obliterating the left maxillary vestibular space extending inferiorly to overlap the external oblique ridge and just lateral to the ramus. It was tender on palpation and the mucosa over the swelling appeared normal. The teeth in the maxillary quadrant were tender on percussion; however, no tooth mobility was noted.

CT scan showed a well-defined mass in the infratemporal region with the presence of an impacted tooth in the left maxillary sinus, and displacement of the posterior wall of the maxilla with involvement of the pterygoid plates. Some resorption on the medial side of the ramus was noted.

Incisional biopsy was done and histopathology was suggestive of a benign odontogenic myxoma.

The patient was taken up for surgical excision of the benign tumor under general anesthesia. For access to the infratemporal region, a classical Weber Ferguson's incision, i.e., Subtarsal (Midtarsal), Lateral nasal (rhinotomy), Subnasal lip (Mid philtral), was marked. An incision was made through skin, subcutaneous tissue, underlying muscle, and periosteum. Vestibular component of the W-F incision was continued to the anterior border of the tumor and further continued across the tumor to the retromolar area. Skin flap was reflected in the subperiosteal plane to expose the left hemimaxilla and infratemporal region. The tumor was found to be well encapsulated and was separated from the surrounding tissues except for the medial surface of the

coronoid process for which an osteotomy was made at the base of the coronoid process and the coronoid process was retrieved attached to the tumor mass (Fig. 2). Layerwise closure was done. Histopathological report showed loose connective myxoid tissue with occasional fibroblast and blood vessel and was thus confirmatory for an odontogenic myxoma. There have been no clinical or radiological signs of recurrence over the past 2-year follow-up.

3. Discussion

Central odontogenic myxoma is a nonencapsulated benign tumor of the jaws that has a rare occurrence. These lesions may be infiltrative, aggressive, and may be recurrent. Peripheral myxoma is slow growing and less aggressive and has a lower recurrence potential as compared to the central myxoma.

The origin of OM is believed to be from the mesenchyme of a developing tooth or the periodontal ligament. Based on available literature, it is still doubtful whether OM is an odontogenic neoplasm. However, its histological similarity to the stellate reticulum of the developing tooth, its exclusive occurrence in close proximity to the tooth-bearing parts of the jaws, the occasional association with the missing or an unerupted tooth, the presence of odontogenic epithelium in minority of cases, and the fact that it rarely appears in other parts of the skeleton are supportive of an odontogenic origin.⁴ Peripheral odontogenic tumors demonstrate histological characteristics of their intraosseous counterparts, but occur solely within the soft tissue covering the tooth-bearing portion of the mandible and maxilla.

Peripheral odontogenic myxoma is rare with less than 6 cases manifesting in the gingival soft tissue as reported in literature. In our present case, the patient was a male

Fig. 2 – (a) Intraoperative exposure of tumor via Weber-Ferguson incision showing tumor arising from left pterygomaxillary area. Tumor mass was found to be well encapsulated. (b) Excised tumor specimen found to have attachment to the left coronoid process.

child, 11 years of age, with a soft tissue myxoma slow growing for over one year. The tumor appeared to be localized in nature, and was very atypically located in the left infratemporal fossa region, that has never before been reported in literature.

The CT scans revealed that it was a well-defined mass compressing the lateral wall of the maxillary sinus, abutting the posterior wall of the maxilla and the pterygoid plates. As it extended anteriorly, it was also causing resorption of the ramus of the mandible. It is worth mentioning that the CT scans showed the presence of an impacted tooth in the maxillary sinus due to its controversial odontogenic origin. On excision, the tumor appeared to be a well-encapsulated lesion and excised in toto. Some soft tissue attachment to the left coronoid process was noted for which left coronoidectomy was done to facilitate easy and complete removal of the tumor mass. But histopathologically, the tumor mass did not seem to arise from the coronoid process. Also the impacted tooth seen on the CT scan was well away into the left maxillary sinus. All the above findings were suggestive of the tumor being an extraosseous, soft tissue myxoma atypically located in the infratemporal fossa region solely associated with the tooth-bearing portion of the jaws.

Surgical excision is best suggested for OM as they are radioresistant tumors. Moreover, the lack of capsule and its infiltrative behavior is responsible for high recurrence rate when managed conservatively with enucleation and curettage.⁴ Due to this locally aggressive behavior of OM and high recurrence rate, there is need for radical resection. OM bears a high risk of recurrence, and mainly due to the gelatinous aspect and the fact that it does not have a capsule.⁵⁻⁷ However, soft tissue, extraosseous myxoma is not reported to cause a recurrence, and a carefully planned conservative approach is justified in these cases. In the present case, it appeared as locally aggressive and invasive on CTs, during surgery it turned out to be a well-encapsulated tumor, and it was easily

removed completely in toto. Also considering the recurrence rate and slight soft tissue attachment to the coronoid process, left coronoidectomy was done. However histological report confirmed no significant association of the tumor with the coronoid process, as well as with the impacted tooth in the maxillary sinus.

A follow-up period is clearly necessary. It has been recommended that patients should be followed closely for at least the first 2 years after surgery, which represents the period during which the neoplasm is most likely to recur.⁸ Our patient has been following up since the past 2 years and there have been no clinical or radiological signs of recurrence.

Conflicts of interest

The authors have none to declare.

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