

Epiphora: A Rare Presentation of Maxillary Odontogenic Myxoma

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Citation

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Abstract

Odontogenic myxomas are tumours derived from mesenchymal elements of dental anlage. We report a unique case of maxillary odontogenic myxoma, with epiphora as the sole presentation. A review of the literature regarding the nature of the tumour, its clinical presentations, histological appearance and treatment modalities is also presented.

BACKGROUND

Odontogenic myxomas are tumours derived from mesenchymal elements of dental anlage.¹ Myxoma of the jaws has been classified as a benign odontogenic tumour composed of odontogenic ectomesenchyme, with or without, included odontogenic epithelium. The evidence for its odontogenic origin stems from its almost exclusive location in the tooth bearing areas of the jaws, its occasional association with missing or un-erupted teeth and the presence of odontogenic epithelium in a minority of cases.^{2,3} In 1958, Zimmerman and Dahlin reviewed 2276 primary bone neoplasms and found only 26 myxomatous jaw tumours.⁴ In 1973, Ghosh et al. reported 10 odontogenic myxomas among 8723 primary bone neoplasms.⁵ In 1975, Fu and Perzin reported 256 non-epithelial tumours of the nasal cavity; Para nasal sinuses and nasopharynx – of these, only 6 were myxomas.⁶

We report a unique case of maxillary odontogenic myxoma, which presented with epiphora. A detailed review of the literature is also presented.

CASE REPORT

A 58 years old Caucasian presented to the Ophthalmology department with a right-sided epiphora for 18 months. A CT-DCG (Dacrocystogram) was arranged by the ophthalmologist.

The scan revealed a mass causing substantial expansion of the right maxillary sinus involving all its borders particularly the superior and the medial walls. The mass also obliterated the right middle meatus and the right nasolacrimal duct.

There was a degree of bony destruction supero-medially and supero-laterally. The lesion was likely to represent a benign mucocele or polyp but a malignant process couldn't be excluded (Figure 1).

Figure 1

Figure 1: Coronal CT scan showing the myxoma in the right maxillary. The mass obliterates the right middle meatus and the right nasolacrimal duct.



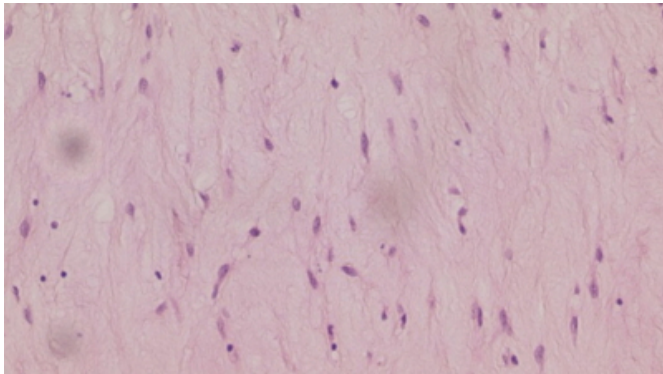
The patient was referred to the ENT department. Nasal endoscopy revealed a greyish polyp originating from the lower part of the right middle meatus. A biopsy was taken from the lesion under general anaesthesia for histological examination.

Microscopically, the sections showed fragments of normal mucosa and fragments of myxoid tissue situated between the surface epithelium and the underlying bony trabeculae. The myxoid tissue consisted of spindle and stellate cells with

small round nuclei, no signs of malignancy were detected (Figure 2).

Figure 2

Figure 2: Histological section of the myxoid tissue consisting of spindle and stellate cells with small rounded nuclei.



Immuno-cytochemical labelling for the stellate and the spindle cells was positive for Vimentin and negative for S100 protein, Desmin, Smooth muscle actin and Cytokeratins (MN116, CAM 5.2). The appearance was highly suggestive of Odontogenic Myxoma.

The lesion was removed via a mid-facial degloving medial maxillectomy approach. The right maxillary antrum was opened through the anterior wall and the medial wall of the maxilla was removed, the tumour was shelled out from the cavity (Figure 3). The ethmoidal and sphenoidal sinuses were explored and there was no obvious tumour in both, the maxillary cavity, the maxillary cavity was packed with white head's varnish on ribbon gauze. En block excision was achieved intra-operatively as there was no evidence of invasion to the floor of the orbit and the lamina papyracea.

Figure 3

Figure 3: Post-operative specimen showing the myxoma completely excised.



Post-operatively the patient made an uneventful recovery. The Whitehead's varnish pack was removed 3 weeks later under G.A, with no evidence of a residual tumour. Complete excision of odontogenic myxoma was confirmed with the histological examination of the tumour specimen.

DISCUSSION

Myxomas usually present as a painless facial swellings, however, tumours of the maxilla tend to enlarge and often fill the maxillary sinus before presenting as a facial swelling. The destructive nature of the tumour can cause nasal obstruction or ocular changes even palatal swellings have been reported. Myxoma tends to affect the posterior part of the mandible more than the maxilla, in ratio 3:2 (4).

The age distribution of myxoma ranges from 1-76 with most cases occurring between 25 and 35 years. Sex distribution is nearly equal and there is no predilection for any racial or ethnic group (7).

Radiologically, odontogenic myxoma commonly shows multiple radiolucent areas of varying size separated by straight or curved bony septa (soap-bubble appearance) (8,9). This appearance may be indistinguishable from that of an ameloblastoma. CT images of the odontogenic myxoma shows osteolytic expansile lesions with mild enhancement of the solid portion of the mass with expansion and thinning of the surrounding bony boundaries. The characteristic finding on CT scan may be the strands of fine lacelike density (10).

Histologically, the tumour cells are small spindle shaped or stellate cells embedded in myxoid background, atypia and

mitosis are rare, vascular invasion has not been reported. However, the tumour shows local infiltration which explains the local recurrence in case of incomplete excision. Immunohistochemically, the tumour cells are positive for Vimentin (pan-mesenchymal marker) and Actin. The myxoid background of myxoma makes it necessary to distinguish this tumour from chondromyxoid fibromas, dentigerous cyst, giant cell granuloma, sarcoma, ameloblastoma and inflammatory polyp with stromal cell atypia (6, 11).

Myxomas of the jaw and the antrum are benign, slow-growing expansile tumours. They can act quite aggressively and the local recurrence is quite common. Most authors recommend conservative surgical excision in the form of local enucleation and curettage for small lesions followed by close follow up and more radical surgery for aggressive disease. Recurrence rates as high as one third has been reported if the surgical margins are not clear. The neoplasm recurs typically during the first two years after treatment, and this is the period during which the patient should be followed closely (12,13). The treatment of choice has traditionally been surgery because myxoma is radio-resistant. Adjuvant radiotherapy is generally not recommended in the treatment of odontogenic myxoma, but some authors advocate pre-operative radiation in order to achieve shrinkage of the tumour. However, benefit from such therapy is not generally accepted (14,15,16).

CONCLUSION

Odontogenic myxoma is a rare maxillary tumour that may present with epiphora.

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