Peripheral Odontogenic Myxoma of Zygoma and Orbital Region - A Unique Case Report with Review of Literature

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Abstract

Rationale: Peripheral odontogenic myxoma (POM) is a rare mesenchymal tumour and it is the first case report of POM involving orbital and zygoma region. **Patient Concerns:** A 16-year-old male presented with a painless, slow-growing swelling over his left infratemporal region. **Diagnosis:** The histopathological examination of the tumour was diagnosed as POM. **Treatment:** The patient was treated by surgical removal of tumour under general anaesthesia. **Outcomes:** The patient has been under follow-up for the past 2.5 years and there has been no recurrence. **Take-away Lessons:** POM is a rare mesenchymal tumour. To our knowledge, this is only the second report of a POM of the infratemporal region and the first report of a myxoma, which extends into the zygomatic region and lateral wall of the orbit.

Keywords: Myxoma, odontogenic tumour, zygoma, orbit

Introduction

Pure myxoma is a very unusual condition. It usually manifests as fibromyxoma, lipomyxoma, odontogenic fibromyxoma.[1] Myxoma and myxofibroma are terms that the World Health Organization uses interchangeably.[2] Myxoma is limited to the jaw bones and accounts for 3%–6% of odontogenic tumours.[3] Only few cases of peripheral odontogenic myxoma (POM) have been reported in the literature, with the mandible as the most frequently affected jaw bone. A case report of a mixed form of POM, which is a rare entity in the zygoma and infratemporal areas, is presented in this paper.

Case Report

A 16-year-old male presented with a slow-growing, painless swelling over his left temporal region for the past 2 months. The patient was asymptomatic and the skin over the swelling was normal [Figure 1]. There was neither paraesthesia nor trauma history. On clinical examination, a widespread, firm, non-tender swelling of approximately 3 cm × 2 cm × 2 cm was observed with mild facial asymmetry. There was no growth observed intraorally.

A subsequent computed tomography scan revealed a distinct, irregularly marginated bony projection arising from the zygoma and extending posteriorly till the infratemporal fossa [Figure 2]. The tumour had a thick cartilaginous capsule. The tumour was roughly 3.5 cm × 2.4 cm × 2.6 cm in size.

A provisional diagnosis of ameloblastoma was made. Differential diagnoses were established for osteosarcoma, fibromyxoid sarcoma, myxoid chondrosarcoma, rhabdomyosarcoma, fibromyxoma and central giant cell granuloma. Under local anaesthesia, an incisional biopsy was carried out, and the histopathological report suggested fibromyxoma.

The tumour was surgically removed under general anaesthesia. The tumour was approached using Keen's approach and the Alkayat Bramley incision intraorally and extraorally, respectively [Figure 3]. Intraoperatively, it became apparent that the encapsulated tumour was extending from the medial surface of the zygoma, extending superiorly to the left lateral orbital rim and inferiorly 0.5 cm below the zygomatic arch.

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Figure 1: A 16-year-old male presented with slow-growing painless swelling of the left temporal region

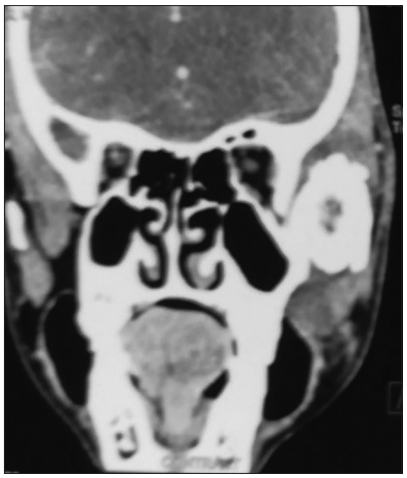

Figure 2: Coronal computed tomography scan showing a mixed radiolucent and radiopaque tumour present in left zygomatic region

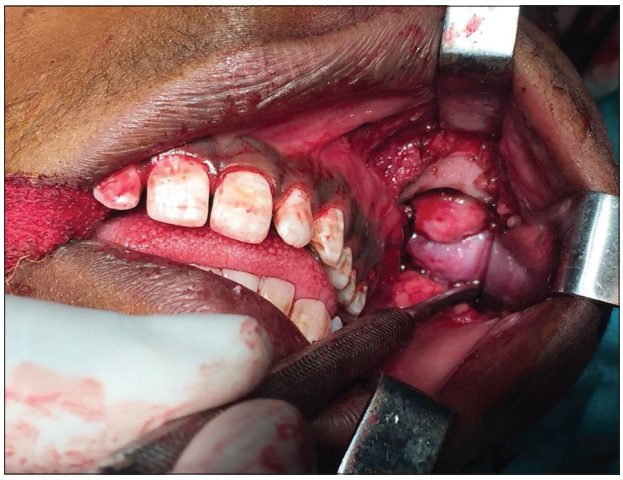

Figure 3: Intraoperative view showing bone resection to expose the tumour present in the left zygomatic region

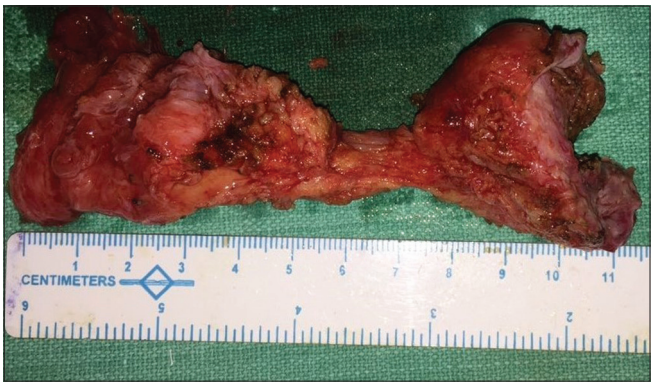

Figure 4: Gross specimen after excision of tumour

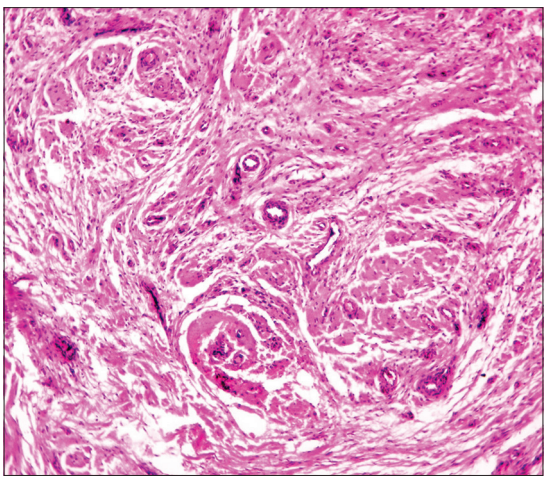

Figure 5: Histopathological image of excised tumour (×10 view)

There was no erosion of the bone. Although the tumour was close to the lateral wall of the orbit and zygoma, the bone remained unaffected and did not exhibit any tumour infiltration.

To preserve the vital structure, a plan for surgical removal of tumour was devised, which included the removal of the tumour in toto with free margins [Figure 4]. Following surgery, the patient made a full recovery with no complications. The resected specimen was sent for histopathological analysis and it was diagnosed as a fibromyxoma. For the past 2.5 years, there has been no radiological or clinical signs of recurrence, and the patient is under regular follow-up.

Discussion

Dutz and Stout claim that Virchow coined the name 'myxoma' in 1863. A particular kind of myxoma known as fibromyxoma has a higher ratio of fibrous/myxoid tissue. Thoma published the first description of fibromyxoma of the jaw in 1947.[1] Based on its origin, myxoma can be categorised as either osteogenic or odontogenic. Odontogenic myxoma is an uncommon benign tumour that frequently invades and recurs locally.

Odontogenic myxoma can be classified into two types: central myxoma and POM, based on their location within or outside of the bone. POM, clinically present as pedunculated or sessile exophytic masses located primarily in gingival POM, is a very rare tumour, and data on its clinical and pathological features remain scarce.

Table 1: Review of literature

Year	Title	Author	Age (year)/ gender	Radiographic examination	Histopathology	Treatment
2008	Maxillary odontogenic myxoma involving the maxillary sinus - case report	Allan *et al*.,	23/female	Osteolytic tumour in the edentulous region and root resorption in the regions of 15 and 17 CT showed a tumour invading the right maxillary sinus, extending throughout the entire wall of the maxillary sinus, extending all the way to the orbit floor	Odontogenic myxoma	Surgical removal of tumour
2010	Odontogenic myxoma: Report of two cases	Sridhar P. Reddy, Ananth Naag, Bina Kashyap	25/male	Periapical radiolucency involving tooth 36, which extends posteriorly	Odontogenic myxoma	Surgical excision with curettage was done under LA
			26/female	Well-demarcated multilocular radiolucent tumour with honeycomb or tennis racquet appearance	Odontogenic myxoma	Radical resection with hemimandibulectomy was advised
2011	POM: A review of the literature and report of two cases	Erich J. Raubenheimer, Claudia E. Noffke	53/female	No bony involvement was present	Odontogenic myxoma	Surgical excision
			38/female	Displacement of the incisors, canines and premolar teeth	Odontogenic myxoma	Surgical excision
2015	Peripheral myxoma of infratemporalregion: An unusual case report	Kapoor P, Gandhewar TM, Andrade NN, Desai RS	11/male	Well-defined mass in infratemporal region with the presence of an impacted tooth in the left maxillary sinus	Odontogenic myxoma	Surgical excision under GA
2015	Odontogenic myxoma: Report of three cases and retrospective review of literature in the Indian population	Chaudhary Z, Sharma P, Gupta S, Mohanty S, Naithani M, Jain A	7/male	No relevant radiographic findings		Surgical excision with peripheral ostectomy
			50/female	Multilocular radiolucency with tennis racket appearance		Anterior partial maxillectomy under GA
			25/female	Multilocular radiolucency		Curettage with peripheral osteotomy under LA
2016	Peripheral odontogenic myxoma	Tasnime S, Saxena C, Bansal V, Wadhwan V	14/female	No radiographic changes	Peripheral odontogenic myxoma	Excision of tumour with curettage under LA
2017	Odontogenic myxoma: A review with report of an uncommon case with recurrence in the mandible of a teenage male	C. Shivashankara, Madhumati Nidoni, Shrish Patil, KT Shashikala	13/male	Multilocular, radiolucent tumour involving the left angle of the mandible, with a thickened bony rim	Odontogenic myxoma	Surgically excised and curetted
2017	POM in 12 years old girl: A rare entity	Kanitkar S, Kamat M, Tamagond S, Varekar A, Datar U	12/female	Drifting of 31 and 32 without erosion of alveolar bone	Peripheral odontogenic myxoma	Excision of tumour with curettage under LA
2020	The rare radiographic sunburst appearance of odontogenic myxomas: A case report and review of the literature	Jamie A. White, Naomi Ramer, Todd R. Wentland, Molly Cohen	34/male	A sunburst appearance of the left posterior mandible	Odontogenic myxoma	Segmental resection of the mandible
2021	Odontogenic myxoma with pain and uncommon histological feature in the mandible: A case report and review of the literature	Armaghan Tarjan, Mostafa Rezaee, Hossein Danesteh, Nazafarin Samirani-Nezhad	52/male	Multinodular radiolucent tumour extending from the apical and distal root of the third molar to the mesial side of the apex of the canine tooth on the right lingual side, with mild root resorption	Odontogenic myxoma	Segmental resection under GA

GA: General anaesthesia, LA: Local anaesthesia, CT: Computed tomography, POM: Peripheral odontogenic myxoma

POM also has a variable radiographic presentation of the tumour. Usually, POM is present with no radiographic changes,[4] but multilocular[5] and sunburst[6] types of radiographic representation are also documented in the literature. POM in young people[5] is rare, and fewer cases were reported in the past. POM is mostly reported in the second and fourth decades of life,[2] with a predilection for the female gender[7] and mandible.[8] In our case report, it was found in a 16-year-old male. The radiopaque type of this tumour usually presents in either of the jaws, but in some cases, it also presents extragnathically.

The microscopic assessment of these types of tumour is necessary to rule out the other possible diagnosis such as central giant cell granuloma, osteosarcoma, fibromyxoid sarcoma, myxoid chondrosarcoma and rhabdomyosarcoma. Clinical representation of these types of tumour can be misdiagnosed as fibroma or ameloblastoma, both of which have a different treatment modality.

The treatment for this type of tumour is either surgical excision or curettage[9] and it varies according to the location or presentation of the tumour. However, some cases reported in the past involved extensive surgical treatment.[10] Due to the unusual site and extension of the tumour, local excision and sparing of the uninvolved structure have been planned for the preservation of function.

Microscopic examination of the tissue sections showed numerous spindle and round-shaped cells [Figure 5] in a mucoid-rich stroma with inactive rests of odontogenic epithelium. The collagen fibrils were sparse, and there were a few small and large budding blood vessels. These features suggest peripheral or extraosseous odontogenic myxoma.

Radiographic examination showed mixed radiolucent and radiopaque mass medial to the zygoma with clear circumcised margins and without any infiltration into the adjacent bone. From these findings, it was concluded that the tumour was extraosseous in nature, and it was diagnosed as POM.

Table 1 compares the radiographic presentation of the tumour, the location of the myxoma and their surgical management. From the table, it is evident that 20% of the cases have no significant radiographic changes at the site of the tumour, and 35% of cases have multilocular radiolucency. In most cases, surgical excision is the treatment modality that is followed for this type of tumour.

Conclusion

The choice of surgical treatment for odontogenic myxoma depends on various factors, such as the location of the tumour, type (primary or recurrent), age, general medical condition and aesthetic need. Conservative surgery has various advantages when compared to radical approaches, such as reduced morbidity and reduced disturbance of facial growth. Considering the surgical treatment, there should be a balance between a successful resection or excision and the maintenance of function.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initials will not be published and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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