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Case Report

Recurrent complex ameloblastoma of infratemporal fossa in a young adult: a challenging case report

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ABSTRACT

An uncommon, histologically benign, but aggressive, tumor of the jaws, ameloblastoma develops from odontogenic epithelium and has the capacity to extensively destroy jaw bones and infiltrate surrounding tissues. After incomplete treatment, recurrences are frequent. The orbit, anterior cerebral cavity, temporal and infratemporal fossa, and other challenging locations can experience recurrences. We report a case of recurrent complex cystic ameloblastoma over the right side of the face in a 31-year-old gentleman who presented with right facial swelling, serous discharge, intermittent pain, and decreased mouth opening for 2 years. Computed tomography (CT) head and neck with 3D reconstruction showed a complex solid-cystic multiloculated mass lesion in the right infratemporal fossa with large cystic components, septations, solid enhancing components, and peripheral calcifications, closely abutting the parotid gland, pterygoids, mandible, and maxillary antrum. Needful evaluation done. The patient underwent wide local excision of the tumor, right infrastructure maxillectomy, superficial parotidectomy, modified radical neck dissection, and microvascular alanine aminotransferase (ALT) free flap reconstruction. Complete surgical resection with negative margins is the hallmark for curative resection. Local recurrences are rare and usually amenable to re-resection. Postoperative radiotherapy for patients at increased risk of local recurrence improves local tumor control.

Keywords: Ameloblastoma, Maxillary tumors, Infratemporal tumors, Complex facial tumors, Recurrent maxillary and mandibular tumors, Odontogenic tumor

INTRODUCTION

The ameloblastoma is a benign epithelial odontogenic tumor that arises from proliferating odontogenic epithelium. It can be derived for example from enamel residues, parts of the developing enamel, from the epithelial lining of an odontogenic cyst, or the basal cells of the oral mucosa.[1] Among odontogenic tumors, it is the second most prevalent type. According to the proposed World Health Organization (WHO) classification, ameloblastoma is classified as conventional, unicystic, peripheral and metastasizing types.[2]

The incidence of ameloblastomas in the maxilla has been reported to be particularly low in retrospective clinical research; the incidence rates in the mandible and maxilla are 1:88 and 1:58, respectively.[3] Mutations in genes of the MaPk pathway have been observed in almost 90% of all ameloblastomas, and BRAF V600e is the most common mutation. Comparing the desmoplastic variety of ameloblastoma to other conventional versions of the tumor, the former occurs more frequently in the maxilla and has a about similar frequency in the mandible. Ameloblastomas in this area can grow significantly and spread via the sinuses due to the maxilla's spongy texture, endangering the cranial cavity and orbits.[4]

The aim of this study was to report a rare case of a conventional complex recurrent ameloblastoma in the maxilla of a young individual and to discuss the evaluation and surgical management of this to prevent further recurrence.

CASE REPORT

A 31-year-old gentleman who presented with right facial swelling, serous discharge, intermittent pain, and decreased mouth opening for 2 years. He gave history of previous surgery for similar issue 1year back, for which details were not available (Figure 1). Swelling was insidious in onset and gradually progressive in size (Figure 2).

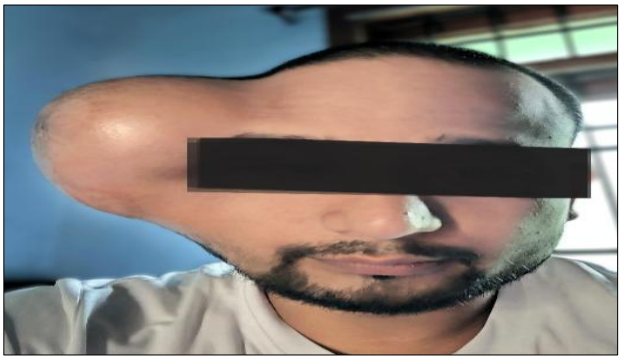

Figure 1: Initial clinical presentation in a 31-year-old male, showing large right facial swelling extending into temporal and infratemporal region.

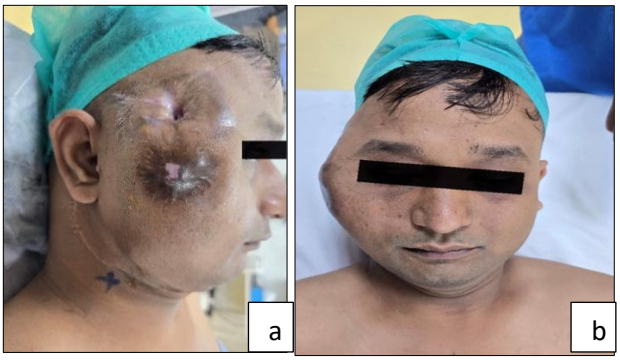

Figure 2 (a and b): Coronal and axial section of contrast enhanced CT head and neck showing complex solid - cystic multiloculated mass lesion in right supratemporal and infratemporal fossa with large cystic components, septations, solid enhancing components and peripheral calcifications.

Patient was planned for contrast enhanced computed tomography (CT) of head and neck with 3D reconstruction, which showed a 6.1×5.7×11.9 cm complex solid - cystic multiloculated mass lesion in right infratemporal fossa with large cystic components, septations, solid enhancing components and peripheral calcifications. The lesion is involving the right masseter muscle, medial pterygoid muscle and zygoma.

Posteriorly indenting the superficial lobe of parotid gland with preserved fat plane. Superiorly, the lesion is extending along the temporalis into the supratemporal fossa. Inferiorly the lesion is extending up to the masseter (Figures 3 and 4).

After discussing in tumor board and surgical excision was planned. After thorough work-up, patient underwent excision of right temporal mass with infrastructure maxillectomy, excision of zygomatic process, right superficial nerve sparing parotidectomy, completion neck dissection. Reconstruction was carried out using the free anterolateral thigh flap to provide bulk in the temporal area (Figure 5).

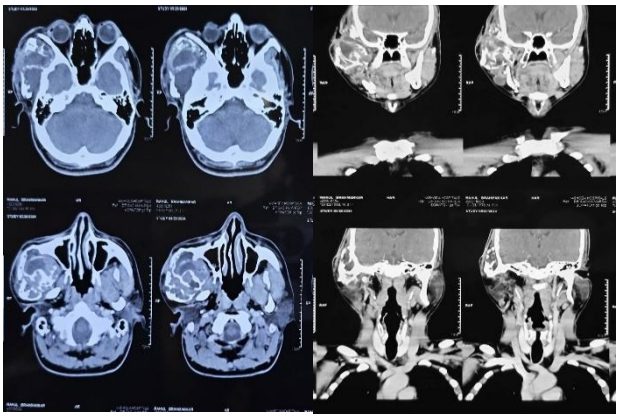

Figure 3: Coronal and axial section of contrast enhanced CT head and neck showing complex solid - cystic multiloculated mass lesion in right supratemporal and infratemporal fossa with large cystic components, septations, solid enhancing components and peripheral calcifications.

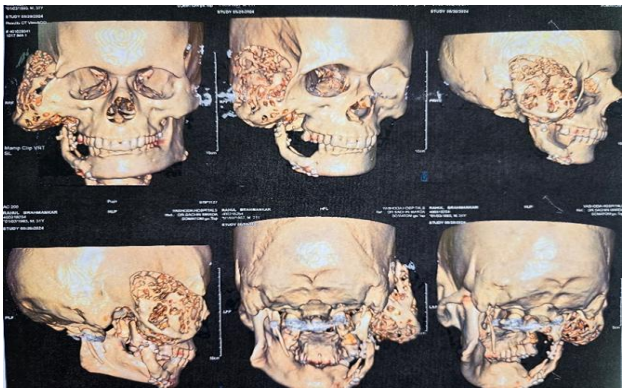

Figure 4: CT neck with 3D reconstruction.

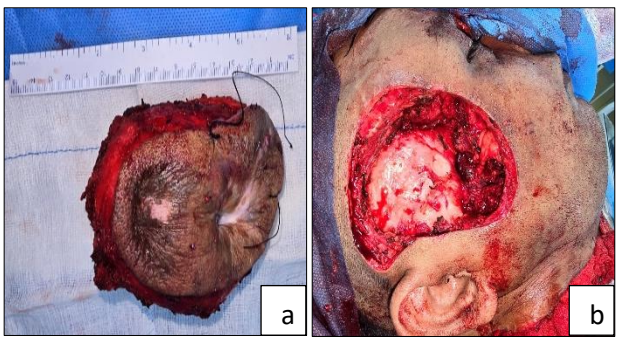

Figure 5 (a and b): Resected specimen and post resection defect over the right side of face.

Histopathological examination shows solid and cystic bony lesion arranged in follicular pattern. Cells show reverse polarity; odontogenic epithelium arranged in nests around gland. Pleomorphic adenoma features in parotid gland. All 17 1ymph nodes are free 0/17 - features are suggestive of recurrent ameloblastoma (Figure 6).

Postoperatively he recovered uneventfully. Patients have been followed up regularly and he remains free of disease clinically and radiographically.

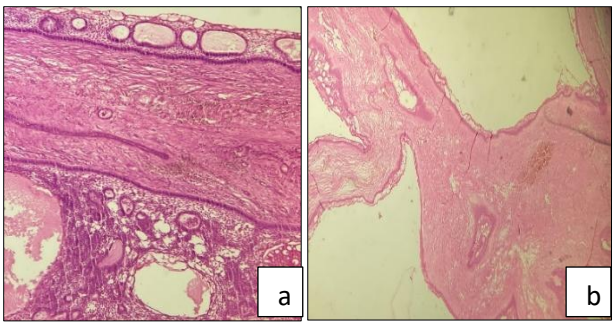

Figure 6 (a and b): H and E Stain showing columnar epithelium with reverse polarity and stellate retinaculum (A-40x) and multiple cysts (B-10x).

DISCUSSION

Ameloblastoma was first recognized by Cuzak in 1927 but Churchill in 1934 was the first one to use the term Ameloblastoma for it.[5] The mandible is often the site of ameloblastoma, which is rather common in young people. Additionally, even in younger patients, the maxillary tumor's proximity to vital anatomical features including the orbit, eyes, sinuses, and air cavities would make the intervention more difficult and need technical and therapeutic talks among the attending physicians. Men and women are equally affected. The mandible to maxilla ratio is 5:1, with the molar region of the mandible being most frequently affected.[6] Maxillary ameloblastomas are less common. It runs an indolent course and therefore remains asymptomatic for a long time and can attain massive sizes. The cosmetic deformity is commonly the presenting complaint.

Ameloblastomas are divided into three clinical categories-solid/multicystic, unicystic, and extraosseous/peripheral. Ameloblastoma exhibits follicular and plexiform features in histology. The follicular type is further classified as acanthomatous, desmoplastic, granular cell, basal cell, clear cell, and mixed variety based on the differentiation.[7] With a comparatively high chance of recurrence, the follicular solid/multicystic (85%), granular cell, and acanthomatous forms necessitate more drastic treatment and careful monitoring. Recurrence probability is comparatively low for the plexiform, unicystic, desmoplastic, and peripheral forms.[8] Recurrence rates are higher with conservative treatments like enucleation and curettage therapy without a safety margin (up to 90% in the mandible and 100% in the maxilla) compared to 5% to 15% with radical resections like en bloc resections with bone margin. Inadequate removal of the affected bone, soft tissues on top, and the spread of leftover bone fragments, seedling of tumor cells carrying tumor tissue could all contribute to recurrence.[9]

Bansal et al advised conservative treatment exclusively to prevent children's functional, psychological, and cosmetic negative effects.[10] In a 5-year follow-up, Antonoglou et al's comparison of conservative versus radical treatment revealed that radical surgery was linked to a lower risk of recurrence.[11] Maxillary ameloblastomas with erosion of bony walls are high risk tumors as they may infiltrate pterygoid muscles posteriorly or extend superiorly into orbital floor, paranasal sinuses. Clinico-radiological follow-up every year is necessary for these high-risk cases.

Given that time is a key role in tumor aggravation and structural loss, the significance of an early diagnosis should be underlined. Professionals must be able to diagnose patients quickly and then decide on the best course of action. Given the need for a thorough assessment of the lesion's internal composition, extent, and shape, the significance of imaging tests becomes clear. This is necessary to plan the intervention as effectively as possible (particularly when the lesion is located in the maxilla) and to foresee issues related to the maxilla's architecture and the biological activity of ameloblastomas.

CONCLUSION

Recurrent complex ameloblastoma of the infratemporal fossa in young adults presents a formidable surgical challenge due to its aggressive local behaviour, proclivity for loco-regional soft tissue and bone invasion, and high recurrence risk even after radical excision with recommended margins. The anatomical complexity of the infratemporal and temporal areas complicates surgical access and definitive removal, often demanding extensive procedures that must be balanced with aesthetic and functional outcomes. Our study concluded that the main contributing factors identified for recurrence were multilocular ameloblastomas, follicular histopathology and conservative treatment methods. Long-term regular follow-up, ideally extending to a decade, is crucial for early detection and management of late and rare recurrences. The case underscores the importance of advanced imaging, multidisciplinary planning, and individualized surgical approaches in optimizing patient prognosis in this rare and challenging presentation.

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