

## **LOCALLY AGGRESSIVE LESION OF THE OROFACIAL REGION : A CASE REPORT**

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### **ABSTRACT**

Ameloblastoma is a benign locally invasive epithelial odontogenic tumour comprising 1% of all tumours and cysts arising in the jaws. It is commonly found in the third and fourth decade in the molar ramus region of the mandible. Ameloblastoma of the lower jaw can progress to variable sizes and cause facial asymmetry, displacement of teeth, malocclusion and pathological fractures. Among all types of ameloblastoma, multicystic ameloblastoma is believed to be locally aggressive lesion that has the tendency for recurrence. In this report we present a large multicystic ameloblastoma in the left body-ramus region of the mandible in a 19-year-old female patient. This large lesion was diagnosed with the help of CT and was successfully managed by hemimandibulectomy with simultaneous reconstruction using iliac crest bone.

**KEY WORDS:** Hemimandibulectomy, multicystic, aggressive lesion

## INTRODUCTION

Ameloblastoma, previously known as Adamantinoma, is a benign locally invasive odontogenic tumour of epithelial origin arising in the orofacial region, significantly in the molar ramus region of the mandible. It is commonly found in the population of third and fourth decade. [1] [2] Multicystic type of ameloblastoma has been proved to have a greater tendency for recurrence. [2] [3] In this report we present an invasive multicystic ameloblastoma of the left body-ramus region of the mandible in a 19-year-old female patient which was diagnosed with the help of CT and was successfully managed by hemimandibulectomy with simultaneous reconstruction using iliac crest bone.

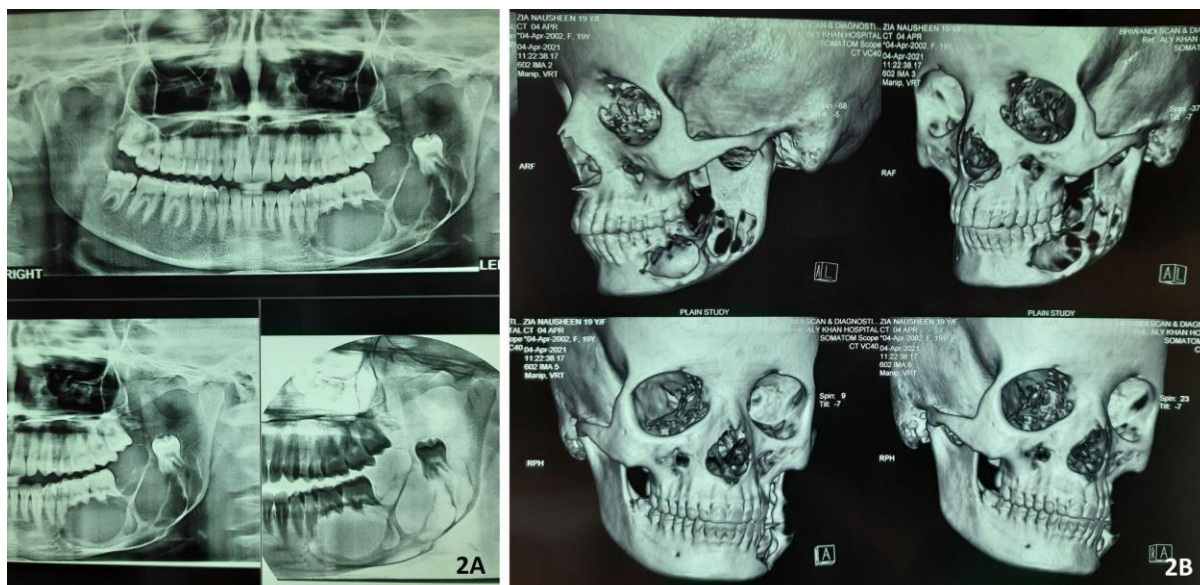
## CASE REPORT:

A 19-year-old female patient reported to the oral pathology and microbiology department of our institution with a slow growing, painless swelling on the lower left side of the face since a year. [Figure 1] The patient was non syndromic, and her medical history was unremarkable, and the interrogation reports showed no previous history of extraction or infectious episode. Extraoral examination revealed a firm swelling distal to the ala of the nose to 2 cms away from the corner of the lips. Intraoral examination revealed a diffuse hard swelling of  $4 \times 3 \times 2.2$  cm in diameter with obliteration of the buccal vestibule extending from the distal aspect of the left mandibular first premolar to the left retromolar trigone region.

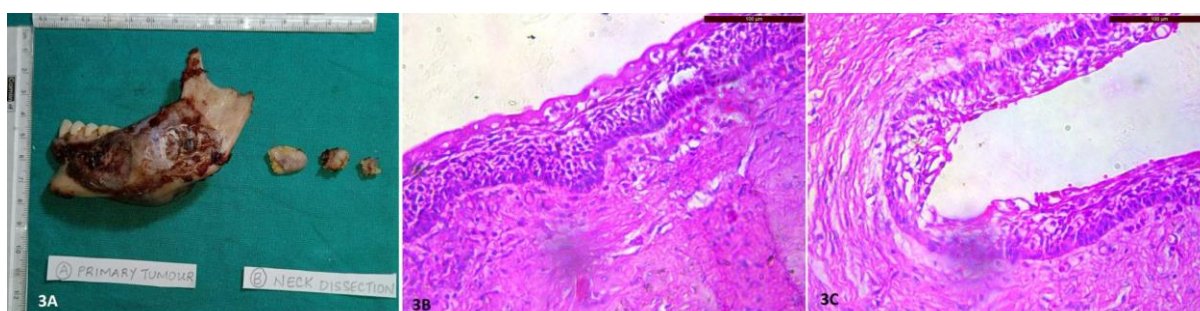


**FIGURE 1:** Clinical photograph shows lesion extending from the distal aspect of the left mandibular first premolar to the left retromolar trigone region.

Panoramic radiograph and computerised tomography revealed a large, lytic, expansile multiloculated lesion from the left inferior alveolar arch and left ramus of the mandible with postero-superior displacement of the left lower third molar. [Figure 2A, 2B]



**FIGURE 2A, 2B:** Panoramic radiograph and computerised tomography showing a large, lytic, expansile multiloculated lesion from the left inferior alveolar arch and left ramus of the mandible with postero-superior displacement of the left lower third molar. Aspiration yielded a yellowish straw-coloured fluid which showed cholesterol crystals on the wet mount, and this was consistent with the diagnosis of a cystic lesion.



**FIGURE 3A:** Figure showing the excised tissue specimen ; **3B, 3C:** Photomicrograph of the excised tissue specimen showing a cystic capsule lined by epithelium consisting of tall columnar cells and an overlying stellate reticulum in a plexiform pattern with connective tissue entrapments. (H & E, 400 x)

H & E-stained soft tissue section of the incisional biopsy showed a cystic capsule lined by epithelium consisting of tall columnar cells and overlying stellate reticulum. Cystic epithelium is seen proliferating luminally and in the underlying stroma in a plexiform pattern with connective tissue entrapments. Large number of blood vessels is seen in the connective tissue entrapments. The overall features were suggestive of Ameloblastoma. Hemimandibulectomy was done with simultaneous reconstruction using iliac crest bone. The histopathological examination of the excised tissue confirmed the preliminary diagnosis. [Figure 3A, 3B, 3C]

## DISCUSSION:

Ameloblastoma is a benign epithelial odontogenic tumour often aggressive and destructive with the capacity to erode bone and invade adjacent structures. Ameloblastoma of the lower

jaw can progress to variable sizes (1–16 cm) and cause facial asymmetry, displacement of teeth, malocclusion and pathological fractures. [1] [4] Ameloblastoma is found in higher frequency in women than men and majority (70%) are located in the molar ramus region and 10–15% are found in association with an unerupted tooth, similar to our case. [1] [5] CT images usually show an expansile, radiolucent, multiloculated cystic lesion, with a characteristic “soap bubble-like” appearance. Other CT findings also include cystic areas of low attenuation with scattered attenuating regions, representing soft-tissue components. [3] Thinning and expansion of the cortical plate with erosion through the cortex can be seen. The associated unerupted tooth may be displaced and resorption of the roots of adjacent teeth is common which is similar to the radiographic picture of our patient. It is worth noting that the desmoplastic variant of the conventional ameloblastoma usually appears as a mixed radiolucent and radiodense lesion, often resembling a benign fibro-osseous lesion, and is most commonly found in the anterior maxilla. [3] Radiographically, ameloblastoma appears either unilocular or multilocular and, histologically, as unicystic or multicystic. Both forms have been shown to recur, particularly following inadequate surgical treatment. [6] The periphery of the lesion may be smooth or scalloped. The cortical plate may become thin, expanded, and may even be perforated if the lesion is in its advanced stage. An occlusal radiograph may demonstrate cyst-like expansion, with thinning of the adjacent cortical plate leaving only a thin 'eggshell' of bone. [6] [7] The benign ameloblastoma includes Follicular, Plexiform, Desmoplastic, Unicystic, Granular, Acanthomatous and Basal cell type. [8] The Follicular ameloblastoma is the most encountered variant which is composed of many small discrete islands of tumour with a peripheral layer of cuboidal or columnar cells whose nuclei are generally well polarised. These cells strongly resemble ameloblasts or preameloblasts and these enclose a central mass of polyhedral, loosely arranged cells resembling the stellate reticulum. [8] [9] The Plexiform type contains basal cells arranged in anastomosing strands with an inconspicuous stellate reticulum. The stroma is usually delicate, often with cyst like degeneration. [8] [9] In the Acanthomatous ameloblastoma, the position of the stellate reticulum undergoes squamous metaplasia, sometimes with keratin formation in the central portion of the tumour islands. This usually occurs in the follicular type of ameloblastoma. On occasion, epithelial or keratin pearls may even be observed. [9] In the Granular cell ameloblastoma, there is marked transformation of the cytoplasm, usually of the stellate reticulum like cells, so that it takes on a very coarse, granular, eosinophilic appearance. [8] [9] The Basal cell type of ameloblastoma bears considerable resemblance to the basal cell carcinoma of the skin, and are generally arranged in sheets, more so than in the other tumour types. [9] The Unicystic ameloblastoma represents an ameloblastoma variant that on gross examination, and not based on the appearance on the radiograph, presents as a cyst, which is similar to our case. [9] In the Desmoplastic type, the stromal component dominates, compressing the odontogenic epithelial components. [9]

The preferred treatment of the ameloblastoma is wide surgical removal, with the possible exception of the luminal variant of unicystic ameloblastoma for which enucleation may be justified. [8] There are different methods of mandibular reconstruction of large defects with microvascular surgery using donor site from fibula, iliac crest, scapula and radial forearm. [1] [10]

**CONCLUSION**

In our case, the tumour was treated with hemimandibulectomy as it was quite an extensive lesion involving the body as well as the ramus of the mandible. Hemimandibulectomy, simultaneously with reconstruction using iliac crest bone reduces the morbidity while retaining the aesthetics of the patient.

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