



## BENIGN AMELOBLASTOMA: A CASE REPORT OF SOFT TISSUE RECURRENCE

## Dental Science

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## ABSTRACT

Ameloblastoma is a rare odontogenic neoplasm of the mandible and maxilla, with multiple histologic variants, and high recurrence rates if improperly treated. Unicystic ameloblastomas are a rare variant which usually occur in younger populations. They are characterized by slow growth and being relatively locally aggressive, with the main site of origin being the posterior portion of the mandible.

## KEYWORDS

ameloblastoma, locally aggressive, soft tissue recurrence, tumour spillage

The Ameloblastoma is a benign locally invasive odontogenic tumour of epithelial origin, among the benign facial tumors, it is one of the most aggressive, with high recurrence rate but rarely undergoes malignant transformation<sup>1</sup>. Etymologically, the name Ameloblastoma is derived from the old French word "amel," which means enamel, and the Greek word "blastos," meaning germ or bud. Over time, this tumor has been referred by many different names including "cystosarcoma," "adamantine epithelioma," and "adamantinoma,"<sup>2, 3</sup>

These tumors represent 1% of all tumors of the oral cavity<sup>4</sup> with 99.1% of tumors prevalent in the mandible<sup>5</sup> with a predilection for the posterior mandibular region.<sup>6</sup> most Ameloblastoma cases occur between the 3rd and 5th decades<sup>7, 8</sup> and does not show any sexual predilection. Clinical presentation is usually painless with cortical bone expansion, associated with or without tooth displacement. Ameloblastoma is classified according to clinical and radiographic findings into three main types<sup>9</sup> peripheral, multicystic and unicystic ameloblastoma. Radiographically multicystic appears as radiolucent multilocular "soap bubbles" and unicystic appears as rounded and well defined radiolucent areas.<sup>10</sup> the best treatment option remains controversial. It includes conservative procedures like enucleation followed with or without other adjunctive methods like curettage and liquid nitrogen cryospray or Carnoy's solution or cauterization and radical approach by marginal resection, segmental resection or even total mandibulectomy. The best treatment option depends on factors such as location, size, age and the clinical type of lesion<sup>10</sup>

## CASE REPORT

A 44 year old male patient reported to the department with the complaint of slow growing painless swelling in the right side of his face since 5 months (figure 1). He was diagnosed of ameloblastoma of right mandible 2 years back for which Condylar disarticulation with segmental resection till 43 was performed and reconstructed using Titanium plates. Plates was removed following an infection 6 months back. Palpation revealed a diffuse firm, hard, non-tender and well defined swelling on the right side extending from temporal region to inferior border of resected mandible measuring 12\*8 cms was noted. No bleeding, pus discharge or sinus were noted. There wasn't any palpable nodes too.

The routine radiographic evaluation failed in determining the extend of lesion hence MRI was taken which revealed well defined thick walled cystic lesion measuring 10.5\*8.3\*8 cm in the right side of cheek extending anteriorly up to the posterolateral wall of right maxillary sinus causing scalloping (figure 2). Postero medially the lesion is seen displacing the right Para pharyngeal fat and lateral pterygoid and laterally causing expansion and thinning of ipsilateral zygomatic arch. Dural tear was suspected posteromedially. A suspicion of soft tissue recurrence of ameloblastoma was made.



Figure 1: Pre-operative extra oral frontal view

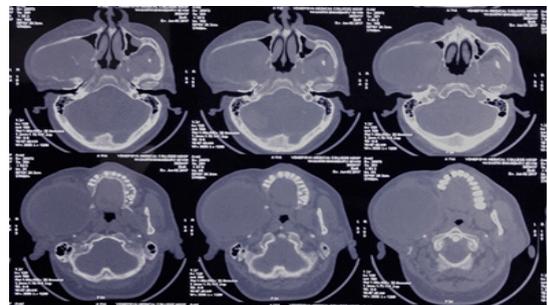


Figure 2 : preoperative MRI axial section showing the extend of lesion.



Figure 3: Gross specimen of the cystic lining

Surgical exploration and wide excision was planned under GA after the assessment of patient's general health condition. The approach was made through the existing scar line. Cyst exposed and the greenish brown content was removed. Cystic lining was separated from the surrounding tissue. Cyst was enucleated in toto (figure 3) and the accessions were sutured in layers. The patient recovered uneventfully without immediate or late postoperative complications. No recurrence noted during the followed up period. Histopathological report showed cystic lesion exhibiting intracystic papillary projections composed of ameloblastomatous epithelium arranged in follicular pattern within a scant fibrous stroma. Basal cells of those islands exhibited peripheral palisading. The central cells were loosely arranged and at places resembled stellate reticulum. Extensive areas of squamous differentiation was also noted. The nuclei were ovoid with granular chromatin and conspicuous nucleoli. Perineural invasion was not seen. They gave an impression of acanthomatous ameloblastoma unicystic type.



Figure 4: Postoperative extra oral photograph

### Discussion

Unicystic ameloblastoma was first described by Robinson and Martinez in 1977<sup>11</sup>. It accounts for 15% of all intraosseous ameloblastomas, with most of the cases occurring in the second decade of life and have a slight male predilection<sup>12</sup>. Approximately 50 to 80% of cases are associated with an impacted or unerupted tooth<sup>13</sup> which leads to the misdiagnosis of odontogenic keratocyst.

The World Health Organization (WHO) has classified ameloblastomas into solid/multicystic ameloblastoma, unicystic ameloblastoma, desmoplastic ameloblastoma and peripheral ameloblastoma, as well as malignant counterparts such as malignant ameloblastoma and ameloblastic carcinoma<sup>14</sup>. The solid/multicystic ameloblastomas are the most aggressive one as they carry a high risk of local infiltration hence more chances of recurrences after inadequate treatment<sup>15, 16</sup>. The unicystic ameloblastoma though they reach large size<sup>17</sup> are less aggressive and is subdivided into 3 groups Luminal, intraluminal and mural. They differ histologically but present clinically and radiographically as unilocular lesions. The most aggressive among the three is the mural variety, in this the epithelium penetrates and breaches the fibrous wall with a high potential to invade the adjacent cancellous bone<sup>18</sup>. Li et al reported a higher recurrence rate of 35.7% for the mural type and a lower recurrence rate of 6.7% for the luminal and intraluminal types<sup>19</sup>.

The anatomic extent of the tumor needs to be carefully assessed by Computed tomography and magnetic resonance imaging.

Several factors influence the rate of recurrence. The first, and probably most important factor, is the clinicopathologic variant of the tumor. There are 3 variants of the benign ameloblastoma, namely solid or multicystic, unicystic, and peripheral<sup>20</sup>. The solid variety has the highest potential for recurrence<sup>21</sup>.

Anatomic site is the second factor that should be considered. 95% of ameloblastomas occur in the mandible<sup>22</sup>. Tumors in the maxilla are far less common but if occur they spread extensively<sup>23</sup>. Hence the recurrence rate of maxillary tumors are 5 times more than mandibular tumours<sup>24</sup>.

A third contributing factor to recurrence is the adequacy of surgery. Due to the locally invasive nature high recurrence rates are reported, the best treatment option remains controversial. It comprises of two surgical options either conservative or radical. Conservative treatment modalities such as curettage, cryotherapy, or enucleation have resulted in recurrence rates of 75% to 90%<sup>25, 26</sup>. These recurrences are very extensive with soft tissue spread.

The "radical" surgical option is the current standard care for ameloblastoma. It is known that ameloblastomas may recur from remaining tumor tissue in the residual stump of the jaw or from adjacent soft tissues<sup>27</sup> hence it requires en bloc resection with 1–2 cm bone margins<sup>28</sup> and reconstruction to restore the function<sup>29</sup>.

There are few studies that even propose radiotherapy for some forms of ameloblastoma. Radiotherapy is advised for inoperable lesions<sup>30</sup> and surgery with radiotherapy<sup>31</sup> and for mandibular recurrences when the first surgery was inadequate; for all recurrences and when the surgical margins are positive after wide resection.

**Conclusion:** Ameloblastoma is a rare, benign, slow-growing but locally invasive neoplasm of odontogenic origin involving the mandible and maxilla. Unicystic variants has high chances of soft tissue recurrences even after radical excision.

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