Recurrent Ameloblastoma 24 Years after Hemimandibulectomy: A Case Report and Review of Literature

Abstract
Ameloblastoma is a benign, locally aggressive epithelial odontogenic tumor with a high recurrence rate. The management of ameloblastoma has always been controversial and an enigma to the surgeons. Literature suggests that 50% of the recurrences occur during first 5 years after the primary surgery, and the recurrence rate following a radical approach such as a segmental resection is 4.6%. The reasons for recurrence after a radical approach can be multifactorial such as remaining stumps, soft tissues, or intraoperative contamination. The purpose of this case report is to emphasize the fact that a recurrence even after 24 years is possible in spite of a radical segmental resection, and hence, a continuous follow-up of the patient is needed and to highlight the fact that the possibility of malignant ameloblastoma or ameloblastic carcinoma should be ruled out when dealing with such ameloblastomas recurring after a long period after a radical primary surgery.

Keywords: Ameloblastoma, radical approach, recurrent

Introduction
Ameloblastoma, a benign odontogenic tumor, was first described by Broca and named by Churchill.[1] It occurs more frequently in the angle and ramus area with no definite sex predilection and mostly diagnosed between the second and sixth decades. The cells of origin of ameloblastoma seem to be epithelial cells that express amelogenin.[2] Although benign, it is locally more aggressive. Ameloblastomas can be classified clinically as unicystic, multicystic/solid, desmoplastic, and peripheral or extraosseous.[3] The intraosseous tumors have high recurrence rate. The management of ameloblastoma has always been controversial and an enigma to the surgeons. Various treatment modalities have been advocated for the management of ameloblastoma ranging from simple enucleation to radical procedures such as resection.[4] However, the factors such as size of the lesion, anatomical location, histological variant, and anatomical involvement should be considered in formulating a treatment modality for the management of ameloblastoma[5] rather a fixed treatment protocol.

In this paper, a case of huge ameloblastoma which has recurred after 24 years even after a radical treatment has been discussed depicting the highly recurrent and aggressive nature of this benign tumor.

Case Report
A 60-year-old male patient was referred to the department of oral and maxillofacial surgery with the complaint of swelling in the lower jaw for 7 months. The history of the patient revealed that he had undergone segmental resection of the left mandible without any reconstruction 24 years earlier. The past surgical reports suggested that it was an ameloblastoma. Extraoral examination revealed a swelling of size 11 cm × 8 cm in the left submandibular region with a segment of mandible missing on the left side. The extensions of the swelling were from the left submental to the angle region with a segment of mandible missing earlier. The past surgical reports suggested that it was an ameloblastoma. Extraoral examination revealed a swelling of size 11 cm × 8 cm in the left submandibular region with a segment of mandible missing on the left side. The extensions of the swelling were from the left submental to the angle region. The consistency was firm, and the swelling was firm in consistency with a normal overlying mucosa. The computed tomography showed a well-defined hypodense lesion in the left submandibular region depicting the highly recurrent and aggressive nature of this benign tumor.
the left mandibular region with a condylar stump remaining [Figure 2b and c]. An incisional biopsy was performed, and the histopathologic report was suggestive of ameloblastoma [Figure 3a].

Surgical excision of the lesion along with the removal of the condylar stump and K-wire was planned under general anesthesia. Risdon’s incison was made on the left side, layerwise dissection was done and the lesion was identified and excised in toto after releasing from the soft tissue adhesions [Figure 4a and b] and the Condylar stump and the K wire were also removed [Figure 5a and b]. The wound was closed layer wise with absorbable sutures. Postoperative recovery of the patient was uneventful, and the specimen was sent for histopathologic examination. The postoperative histopathologic report [Figure 3b] was consistent with the preoperative report [Figure 3a] confirming the diagnosis of ameloblastoma showing follicles of odontogenic islands characterized by peripheral layer of tall columnar cells with and without reversed polarized hyperchromatic nuclei enclosing central cells ranging in appearance from stellate to acanthomatous and granular [Figure 3b]. The patient is disease free after a 3-year follow-up [Figure 6].

Discussion

Ameloblastoma, a benign odontogenic tumor, represents 10% of all jaw tumors. It is most frequently encountered in the mandibular posterior region and usually diagnosed between 30 and 60 years. In the current case, taking into account the site and a history of the treated case of ameloblastoma, a provisional diagnosis of ameloblastoma was made. Despite a clinical and radiographic suspicion in favor of ameloblastoma, a biopsy is always mandatory in such extensive lesions to exclude a malignancy.

The recurrence rate after curettage is 50%–90%. Conventional ameloblastomas tend to infiltrate intact cancellous bone at the periphery of the lesion before bone resorption actually becomes evident clinically and this could be one of the reasons for the recurrence after conservative approaches. The multicystic ameloblastoma has more aggressive behavior and a greater tendency to recur than the unicystic, and hence, it was presumed that the primary lesion could have been a multicystic ameloblastoma.

Radical surgeries such as marginal resection and segmental resection with the inclusion of adjacent soft tissues still have 5%–15% chances of recurrence. The recurrent lesion could arise from the remaining stumps, soft tissues or by intraoperative contamination during the primary surgery or a combination of all the above. In the reported case since the condylar stump was remaining, it could be considered as one of the causes for recurrence.
Al-Bayaty et al[10] and To et al[9] in their individual case reports on recurrent ameloblastoma reported that tumor seeding during surgery could be cited as the cause for recurrence which again could be a possibility in the current case. Literature suggests that 50% of the recurrences occur during first 5 years after the primary surgery.[11] In contrary to this, Mainenti et al.[12] reported a case of recurrent ameloblastoma 33 years after hemimandibulectomy. Siar et al.[13] in their analysis on a retrospective study on 340 cases of ameloblastoma in Malaysian population reported that the mean period of recurrence was 7.3 years with a range of 1–29 years. Hong et al.[14] in a retrospective study of 239 patients with ameloblastoma reported a recurrence rate of 4.5% after segmental resection, 11.6% after marginal resection, and 29.3% after conservative treatment modalities. The present case wherein a recurrence has been reported 24 years after a segmental resection is a clear evidence of the aggressive and recurrent nature of this benign lesion.

Another important aspect to be reviewed when encountering recurrent ameloblastomas is to exclude the possibility of a metastatic malignant ameloblastoma and ameloblastic carcinoma. The lung is the most common site of metastasis (80%).[15] According to Van Dam et al.[16] the average time from the diagnosis of primary to metastasis is 18 years. In the present case, the patient reported with a recurrence after 24 years; hence, this was also taken into consideration, and the patient was subjected to preoperative and the postoperative chest radiographs after 2-year follow-up, and a thoracic opinion was obtained, and no signs metastasis was detected ruling out a malignant ameloblastoma, and histologically, there were no signs of malignancy in favor of an ameloblastic carcinoma.

**Conclusion**

The possibility of an iatrogenic tumor cell implantation during the surgical procedures should be emphasized. An intraoperative seeding of the tumor cells is strongly suspected as the reason for recurrence in the reported case in spite of a radical treatment of the primary lesion. The most important but difficult aspect in the management of ameloblastoma is a close follow-up of the patient because a recurrence even after 24 years is possible despite a radical primary treatment and hence a continuous follow-up, radical approach along with avoidance of intraoperative tumor seeding is required to avoid recurrence in this locally aggressive highly recurrent benign tumor.

### 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 name and initial will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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### Conflicts of interest

There are no conflicts of interest.

### References

1. Reichart PA, Philipsen HP, Sonner S. Ameloblastoma: Biological profile of 3677 cases. Eur J Cancer B Oral Oncol 1995;31B: 86-99.
2. Kumamoto H, Yoshida M, Ooya K. Immunohistochemical detection of amelogenin and cytokeratin 19 in epithelial odontogenic tumors. Oral Dis 2001;7:171-6.
3. Barnes L, Eveson JW, Reichart PA, Sidransky D, editors. World Health Organization Classification of Tumours. Pathology & Genetics. Head and Neck Tumours. World Health Organization. International Agency for Research on Cancer. Lyon: IAC Press; 2005.
4. Müller H, Slootweg PJ. The ameloblastoma, the controversial approach to therapy. J Maxillofac Surg 1985;13:79-84.
5. Sampson DE, Pogrel MA. Management of mandibular ameloblastoma: The clinical basis for a treatment algorithm. J Oral Maxillofac Surg 1999;57:1074-7.
6. Becelli R, Carboni A, Cerulli G, Perugini M, Iannetti G. Mandibular ameloblastoma: Analysis of surgical treatment carried out in 60 patients between 1977 and 1998. J Craniomaxillofac Surg 2002;13:395-400.
7. Pogrel MA, Montes DM. Is there a role for enucleation in the management of ameloblastoma? Int J Oral Maxillofac Surg 2009;38:807-12.
8. To EW, Tsang WM, Pang PC. Recurrent ameloblastoma presenting in the temporal fossa. Am J Otolaryngol 2002;23:105-7.
9. Martins WD, Fávaro DM. Recurrence of an ameloblastoma in an autogenous iliac bone graft. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2004;98:657-9.
10. Al-Bayaty HF, Murti PR, Thomson ER, Niamat J. Soft tissue recurrence of a mandibular ameloblastoma causing facial deformity in the temporal region: Case report. J Oral Maxillofac Surg 2002;60:204-7.
11. Olatan AA, Arole G, Adekeye EO. Recurrent ameloblastoma of the jaws. A follow-up study. Int J Oral Maxillofac Surg 1998;27:456-60.
12. Mainenti P, Oliveira GS, Moraes HM, Blumer Rosa LE. Recurrent ameloblastoma after 33 years of hemimandibulectomy – Case report. Appl Cancer Res 2008;28:37-40.

13. Siar CH, Lau SH, Ng KH. Ameloblastoma of the jaws: A retrospective analysis of 340 cases in a Malaysian population. J Oral Maxillofac Surg 2012;70:608-15.
14. Hong J, Yun PY, Chung IH, Myoung H, Suh JD, Seo BM, et al. Long-term follow up on recurrence of 305 ameloblastoma cases. Int J Oral Maxillofac Surg 2007;36:283-8.
15. Verneuil A, Sapp P, Huang C, Abemayor E. Malignant ameloblastoma: Classification, diagnostic, and therapeutic challenges. Am J Otolaryngol 2002;23:44-8.
16. Van Dam SD, Unni KK, Keller EE. Metastasizing (malignant) ameloblastoma: Review of a unique histopathologic entity and report of Mayo clinic experience. J Oral Maxillofac Surg 2010;68:2962-74.