

Ameloblastoma relapse after 16 years of resection in symphysis of mandible sparing the bone graft

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ABSTRACT

Ameloblastoma is a tumor derived from epithelium involved in odontogenesis. Although it is considered a benign tumor, its clinical behavior may be regarded as lying between benign and malignant. It is characterized by slow but persistent growth, local infiltration into adjacent tissues and recurrences; however, metastases are rare. Diagnosis mainly from tissue biopsy and characteristic finding on plain X-rays does assist in differentiating between types of ameloblastoma. The challenges in the management of this tumor are to provide complete excision as recurrence may occur in incomplete removal and also to reconstruct the bony defect in order to give reasonable cosmetic and functional outcome to the patient.

Key words: Ameloblastoma relapse, epithelial odontogenic tumor, mandibular reconstruction

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INTRODUCTION

Ameloblastoma is slow-growing, locally invasive, odontogenic tumor of the jaw with high recurrence rate if not treated adequately but with virtually no tendency to metastasize.^[1] Ameloblastoma is the second most common odontogenic tumor. It exhibits no sex predilection and occurs over a wide age range. Most cases are diagnosed between 30 and 60 years, whereas the tumor is rare in age younger than 20 years. Geographic and racial differences have been described.^[2,3] Exact etiology of ameloblastoma is not known. Thus, the tumor conceivably may be derived from

- Cell rest of enamel organ, either remnants of dental lamina or remnant of Hertwigs sheath (cell rest of Malassez);
- Epithelium of odontogenic cyst (particularly dentigerous cyst); and
- Disturbances of developing enamel organ.^[4]

The tumor occurs exclusively in the jaw and rarely in the sinonasal cavities. Approximately 80% occur in the mandible, with marked predilection for the posterior region.^[5] This paper presents a case of a man who was operated in 1993, when he was 32 years old, for an ameloblastoma in the left body region of mandible. The surgical treatment was segmental resection of mandible; reconstruction of the defect was performed with a non-vascularised autologous bone graft of the iliac crest after 6 months of resection. After 16 years, ameloblastoma relapsed in the symphysis region without involving the graft.

CASE REPORT

A 32-year-old male patient reported to the Department of Oral and Maxillofacial Surgery in October 1993 with a chief complaint of swelling on the left side of lower jaw. The swelling was hard and had a duration of 6 months. It was gradually progressing in size and was associated with pain and paresthesia for the past 1 month. On I/O- Intraoral examination, the swelling extended from left lateral incisor upto second molar. There was more expansion of buccal cortex than lingual cortex. Expansion had caused the obliteration of buccal vestibule. On radiographic examination, well-defined multilocular radiolucency extending from lateral incisor to second

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Figure 1: Preoperative radiograph of ameloblastoma in November 1993

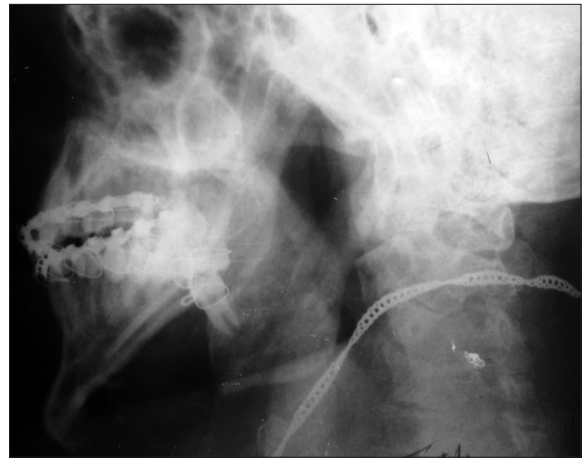


Figure 2: Immediate postoperative (January 1994) after resection

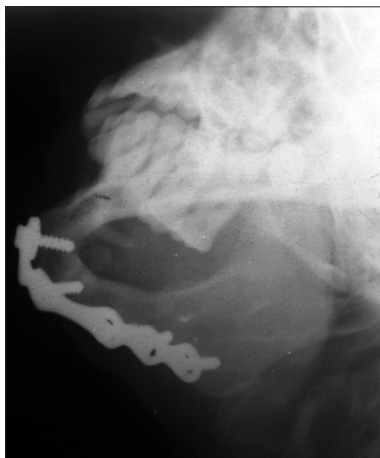


Figure 3: Postoperative (2 years after the reconstruction with iliac crest in 1996)

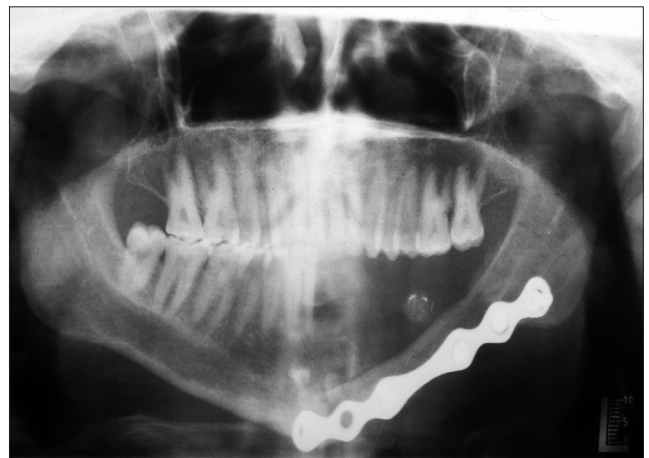


Figure 4: Postoperative OPG in 2006 after 12 years of resection



Figure 5: Postoperative OPG showing relapse in 2008

molar was seen [Figure 1]. Incisional biopsy was done under local anesthesia. On h/p- histopathological examination, ameloblastoma was confirmed. Segmental resection of lesion with 1 cm of normal bone was done [Figure 2]. After 6 months of resection, reconstruction was done with nonvascularized iliac crest bone graft

and stabilized with AO reconstruction plate [Figure 3]. The patient was kept under regular observation for a period of 6 months. In due course of time, mandibular prosthesis was given as replacement for missing teeth. No complications in the postoperative period were reported till 2008 [Figure 4].

In December 2008, the patient reported to the department with a complaint of painless swelling in the labial vestibule, from the past 2 months, in relation to apical region of right lateral incisor and canine teeth. Aspiration revealed 2–3 ml of straw-colored fluid, after which the swelling subsided, which later recurred in 2 weeks. On radiographic examination [orthopantomogram (OPG)], multilocular radiolucency at the level of right symphysis region was seen with no involvement of the bone graft [Figure 5]. Because the lesion was completely contained within the healthy bone, marginal resection of the bone was done under local anesthesia with 1 cm margin [Figure 6]. On microscopic examination, recurrence of ameloblastoma was confirmed. Graft had consolidated markedly well

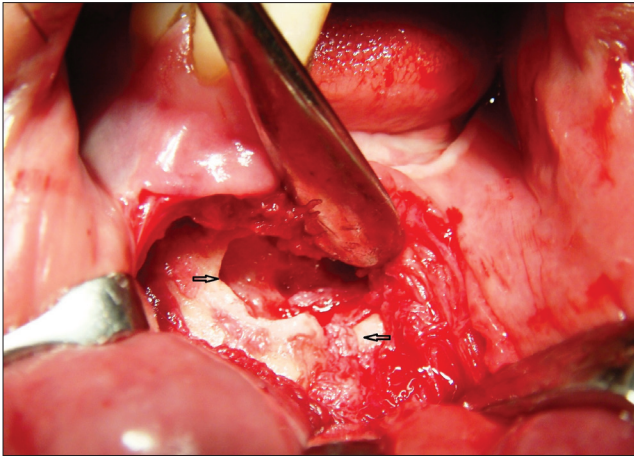


Figure 6: After marginal resection of relapse (right arrow) and butt joint between graft and normal bone (left arrow)



Figure 7: Intraoral view after marginal resection in 2010



Figure 8: After intraoral prosthesis

with normal bone and to our surprise, relapse of the lesion was noticed on normal side and no recurrence was noticed on the bone graft [Figure 7]. The patient was kept under regular follow-up after every 3 months, no recurrence has been reported till date [Figure 8].

DISCUSSION

Ameloblastoma is a tumor with well-known propensity

for recurrence.^[6] Several factors that may influence the rate of recurrence have been identified. The first and the most important is clinicopathologic variant of tumor. It is generally accepted that there are three variants of the benign ameloblastoma, designated as solid or multicystic, unicystic and peripheral.^[7,8] The solid variety has the greatest propensity for local infiltration and therefore the highest potential for recurrence.^[9] The second factor that should be considered is the anatomic site.^[10] Up to 95% of ameloblastomas occur in the mandible. The dense cortical bone of the mandible prevents the tumor from spreading extensively for several years, although spread in the central cancellous bone is beyond the radiographic margins of the tumor.^[11] The third factor contributing to recurrence is the adequacy of surgery. To ensure that lesion is completely removed, the anatomic extent of the tumor needs to be carefully assessed. The lesions that are completely intraosseous can be adequately assessed with standard radiography. Radiologically, the lesions are expansile, with thinning of cortex in the buccal–lingual plane. The lesions are classically multilocular cystic with a “soap bubble” or “honeycomb” appearance. Finally, the histological variant of the ameloblastoma has been suggested to be of prognostic significance in terms of recurrence. Treatment of ameloblastoma varies from enucleating and curettage to *en bloc* resection.^[12] The treatment of choice depends on several factors. Multilocular ameloblastomas have higher recurrence rates than unilocular ones. Age is another important factor when considering the treatment options. The best treatment is still controversial. Since ameloblastomas infiltrate within the cancellous spaces more, the tumor margin goes beyond the apparent clinical and radiographic margin. The attempts to remove the tumor by curettage may leave small tumor islands in bone, which may later occur as recurrences.^[13]

Marginal resection is the most common treatment approach; however, there are reports of 15% recurrence. It minimizes the mandible defect, but can only be used in selected cases. Because of the above factors, segmental resection in our case was done in the initial surgery. Foster *et al*, reported that vascularized bone flap can rebuild any defect extension, whereas bone grafts should have their use restricted to smaller defects that are shorter than 5 cm in length. The successful rate of graft does depend not only upon size, but also upon the contact surface of well-adjusted stumps, well-vascularized receiving bone margins, tight sealing of the oral mucosa, graft stillness with internal rigid fixation and maintenance of satisfactory dental occlusion. In our case, reconstruction was done after 6 months of resection with iliac crest bone graft and stabilized with AO-Osteosynthesis plate.^[14] Removable partial denture was given to the patient to restore the esthetics

and function after 6 months of reconstruction. After a gap of 15 years approximately, recurrence was noticed again but was seen on the other side of the bone; the graft and its junction was absolutely healthy. There are case reports of such recurrences of ameloblastoma after a due course of time, predominantly in grafts and also in the normal bone. But in our case, there was no involvement of graft, with the graft uptake remarkably well with no recurrence.

CONCLUSION

Successful treatment is the one that renders an acceptable prognosis, causing minimal disfigurement and is based on the behavior and potential of the tumor. The result of this study shows that even after segmental resection and reconstruction with iliac crest, chances of recurrence for solid ameloblastoma of mandible are still there. So, more important is that long-term follow-up of at least >10 years for each case of ameloblastoma should be indicated.

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