

Graft Necrosis Occurred After Iliac Crest Reconstruction After Mandibular Segmental Resection of Ameloblastoma

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Abstract: Ameloblastoma is an uncommon benign odontogenic neoplasm of the maxillofacial region constituting less than 1% of tumors of the oral cavity. Ameloblastomas have been categorized broadly into 3 biologic variants: cystic (unicystic), solid, and peripheral. Unicystic ameloblastoma is a rare and less aggressive variant of ameloblastoma. The aim of this report is to describe a case of cystic ameloblastoma treated with segmental resection and iliac graft reconstruction. The possible reasons of graft failure seen in our patient at the early stage of the healing were also discussed.

Key Words: Ameloblastoma, mandible, segmental resection, iliac crest, graft necrosis

Ameloblastoma is a benign, enamel tissue tumor, which does not differentiate to form the enamel. Although considered a benign tumor, its clinical behavior can be considered of middle ground between benign and malignant. According to the WHO 1992 definition, ameloblastoma is a benign but locally invasive polymorphic neoplasm consisting of proliferating odontogenic epithelium, which usually has a follicular or plexiform pattern, lying in a fibrous stroma. The tumor is characterized by slow but persistent growth and infiltration in adjacent tissue.^{1,2} True ameloblastomas are classified into 3 subtypes: solid and multicystic ameloblastomas, unicystic ameloblastomas, and peripheral ameloblastomas.³

Unicystic ameloblastoma was first described by Robinson and Martinez⁴ in 1977 as a special type of ameloblastoma. It has a typical unilocular radiographic appearance and macroscopically cystic nature. It predominantly affects the relatively younger age population, between the ages of 10 and 19 years.⁵ Unicystic ameloblastoma usually appears very similar to a nonneoplastic odontogenic cyst^{6,7} and is frequently clinically misdiagnosed as dentigerous cyst and odon-

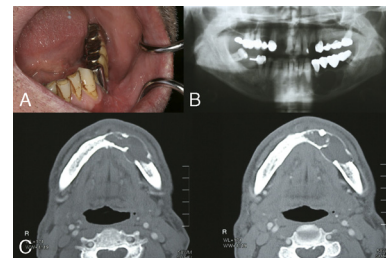


FIGURE 1. A, Intraoral view of the patient. B, Orthopantomographic view of the patient. C, Axial CT image shows the bone expansion.

togenic keratocyst, hence histological confirmation is mandatory. Histologically, the tumor forms the cyst wall and may proliferate into the lumen. Tumor cells forming the cyst wall are flattened and easily mistaken for those of a nonneoplastic cyst.⁵

Various treatment modalities have been used for ameloblastomas including segmental or marginal resection or conservative treatments including enucleation^{4,8-10} and curettage.⁸ Unicystic type of ameloblastoma has a considerably better overall prognosis and much reduced incidence of recurrence compared with conventional ameloblastoma.¹¹

In this report, the diagnosis, treatment, and postoperative complication of a case of unicystic ameloblastoma was presented.

CLINICAL REPORT

A 51-year-old male patient was referred to the Faculty of Dentistry of Ondokuz Mayıs University for prosthetic rehabilitation. The patient's medical history was uneventful except for hepatitis B. No facial asymmetry and lymphadenopathy was observed at the extraoral examination. Intraoral examination revealed an asymptomatic firm buccolingual swelling between the left canine and second molar teeth (Fig. 1A). The panoramic radiograph showed a multilocular radiolucent area with soap-bubble appearance between the right mandibular canine and the left second molar teeth (Fig. 1B). Axial and coronal computed tomography (CT) revealed buccolingual expansion (Fig. 1C). An incisional biopsy was performed and histopathological examination revealed cystic ameloblastoma. Considering the possibility of a recurrence, we planned marginal resection with 1 cm of safety margin and immediate reconstruction with iliac bone graft and titanium reconstruction plate.

Operation was performed under general anesthesia. The lesion was exposed via an intraoral approach. The reconstruction plate was molded in the mandibular arch before resection in order to preserve the facial contour. Segmental resection was performed between the right canine and left 3 molar teeth. Approximately 0.5 cm of basal bone was preserved and a 7-cm-long segment was cut from the mandible in 2 pieces (Fig. 2A). A fracture occurred at the basal bone during the extraction of the segments and the fragments were immobilized

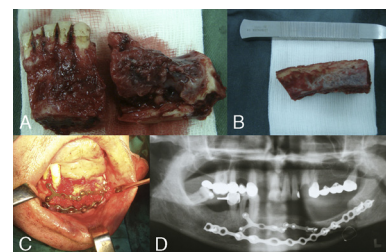


FIGURE 2. A, Extracted segments in 2 pieces. B, Iliac crest bone graft. C, Intraoperative view after reconstruction plate and bone graft placement. D, Orthopantomographic view after the operation.

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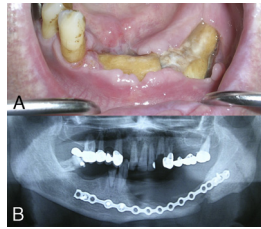


FIGURE 3. A, Intraoral view of the bone exposure. B, Orthopantomographic view after the extraction of the bone graft.

with a single intraosseous wire. Simultaneously with the resection, another surgical team removed an iliac crest bone graft (Fig. 2B). The graft was placed and fitted to the resection site and immobilized by the reconstruction plate with bicortical screws. In order to help keep the graft in position, we installed 2 more of the 1.5-mm system plates with monocortical screws near the mandibular crest (Fig. 2C). The intraoral wound was closed primarily by 3-0 silk suture and iliac crest was closed with 3-0 vicryl and prolene suture. The patient was discharged on the third day of the postoperative period, without any complications. The excisional biopsy result confirmed the first histopathological diagnosis. In his first return, mandibular contour was good, mucosa was intact, and panoramic radiograph revealed good graft position (Fig. 2D). Seven days after surgery, mucosa dehiscence and exposition of the graft were observed. The wound was irrigated daily with povidone iodine and saline solution for 1 month and a chlorhexidine mouthwash was prescribed to the patient. However, dehiscence became wider and the graft was infected (Fig. 3A). One month later, a second operation was performed under local anesthesia to take off the infected graft segment (Fig. 3B). Wound margins were reexcised and closed primarily with 3-0 silk suture. A second exposure occurred 7 days after the second operation. The complete graft was removed 2 months after the second operation. Postoperative second week intraoral examination revealed good mucosal healing (Fig. 4A) and prosthetic rehabilitation was performed successfully (Fig. 4B). Four-year follow-up of the patient was satisfactory.

DISCUSSION

The unicystic ameloblastoma is considered a variant of the solid or multicystic ameloblastoma, accounting for 6% to 15% of all intraosseous ameloblastomas.¹² It occurs in younger patients in the first through third decades, with an average age of 22 years.¹³ Consistent with the literature, our patient was a 51-year-old man. The clinical symptoms of ameloblastoma in the mandible include swelling, dental malocclusion, pain, paresthesia, and anesthesia of the affected side; on the other hand, it could be asymptomatic and diagnosed during routine plain radiographic films.¹⁴ In the present case, a firm buccolingual swelling was observed on intraoral examination. However, the patient was not aware of the swelling and the lesion was detected in the routine dental examination.

Orthopantomograms are useful in the first-line investigation of tumoral and cystic lesions of the jaws. Preoperative CT scan is imperative in enabling the surgeon to make the important decision of whether to curette or to resect. Both bone and soft tissue windows are helpful in evaluating cortical thinning, perforation, and soft tissue involvement. Although magnetic resonance imaging provides better soft tissue imaging, the important relationship of the tumor to bone is more precisely defined by CT.¹⁵

The unicystic ameloblastoma grows predominantly as a cystic lesion. The epithelium lining of the cystic cavity of the neoplasm shows typical cytomorphic features that are recognizable as

ameloblastoma, with a basal cell layer composed of columnar cells displaying hyperchromatic, palisaded nuclei.¹⁶ If the tumor exhibits an infiltrating growth into the wall of the cyst and possibly beyond into the surrounding bone, it is termed as mural (or intramural) growth.¹³ The epithelium may remain in direct contact with the cystic ameloblastic epithelium or it may appear as separate islands of tumor in the connective tissue wall.¹⁷ The presence of a mural component would change the treatment modality if known before surgery. Ackerman et al suggested a more aggressive approach to the treatment of this tumor.¹⁸ Pathologists note that if mural proliferation is present and the lesion has been treated by enucleation or curettage, one never can be certain that the tumor has not penetrated the surrounding bone because the bony margin has not been examined microscopically. In the present case, preoperative histopathological examination after incisional biopsy revealed unicystic ameloblastoma. There was no evidence of mural component so we did not prefer segmental resection. The tumor was excised with safety margins. Postoperative histopathological examination also confirmed the initial diagnosis.

Although ameloblastomas usually progress slowly, they are locally invasive, uncontrolled, and may cause significant morbidity or sometimes death.¹⁹⁻²¹ Patients with ameloblastomas can be treated in many different ways depending on the patient's age, histopathological type, and the localization of the tumor.²² The treatment modality is still controversial and varies from enucleation and curettage to resection.^{1,2} Minimal surgical approach is recommended in children and young patients with ameloblastoma.²³ Maxillary lesions behave distinctly different from mandibular lesions.^{16,24} The higher cancellous bone percentage in the maxilla facilitates the spread of the ameloblastoma, whereas the density of the cortical plates in the mandible tends to limit spread of the neoplasm.^{16,25} Multilocular ameloblastomas have higher recurrence rates than unilocular ones. Conservative treatment of unilocular ameloblastoma with enucleation and/or curettage has an excellent chance of success.^{12,26} Leaving a safety margin of at least 1 cm beyond the tumor radiographic limits is advocated.^{19,27} However, some authors advocate segmentary resection or en bloc resection, which allows total tumor removal and have lower recurrence rates. Multiple recurrences, even with radical retreatment, were also reported.²⁸ The disadvantage of the segmental resection is the resulting facial deformity and function loss if not properly rebuilt.²⁰ The advantage of the marginal resection technique is minimizing the mandibular defect; however, it can only be used in selected cases.²⁷ In the present case, we preferred marginal resection instead of segmental resection, considering the lower recurrence rate of unicystic ameloblastoma. A thin cortical layer was left intact at the lower margin of mandible. However, an unexpected fracture occurred at the marginal bone during the removal of the resected segment. The 2 marginal bone segments were fixed with wire osteosynthesis. No other intraoperative complication occurred.

Immediate reconstruction of mandibular defects avoids obliterated dead space, which may accumulate fluid with increased risk of infection and graft failure, leading to cosmetic disfigurement and difficulties in eating and speech.¹¹ Iliac crest is the most commonly



FIGURE 4. A, Three months after second surgery. B, View of the final prosthesis.

used free graft for mandibular reconstruction. The advantages of iliac crest in mandibular defects are its natural curvature, the abundance of vertical and horizontal height of bone available for mandibular contour, and osseointegration.²⁹ In the present case, the graft harvested from iliac crest was in proper size and shape for the reconstruction and recontouring of the mandible after resection. No donor-site complication was encountered at the postoperative period.

There is no agreement in what sized mandibular defects should be attempted with nonvascularized autogenous bone graft reconstruction.³⁰ Foster et al reported that mandibular segments larger than 5 cm treated with bone grafts tend to have a higher rate of postoperative complications.²⁰ The percentages of the free grafts which need re-exploration were reported to be 2.8% to 20% and the salvage rates of the grafts were 31.8% to 76.1%.²¹ However, in the literature, high success rates were reported with free grafts in which mandibular defects were longer than 5 cm.^{31,32} In this case, the graft placed at the resected area was approximately 7 cm. The reason of the graft exposure in our case was vascular compromise of the graft due to its size. In this case, flap dehiscence and graft exposure were observed at the postoperative seventh day. Instead of early surgical closure, we wait until the soft tissue matures. During the waiting period, the grafted site was irrigated 4 to 5 times a day with chlorhexidine solution to reduce bacterial infiltration. Four weeks later, a second surgery was performed to close the grafted site. The graft volume was reduced, small holes were created at the remaining bone with a round bur, and the soft tissue was resutured. However, a second exposure occurred 7 days after the second operation. The complete graft was removed 2 months after the second operation. Although the graft procedure failed, the reconstruction plate saved the mandibular integrity and we were able to make a prosthesis that provides the patient's function and aesthetics.

CONCLUSIONS

Unicyclic ameloblastoma is a form of ameloblastoma and could be treated with less aggressive treatment modalities than the multicystic variant. Although successful results were reported at reconstruction of mandibular defects with nonvascularized autogenous bone graft, a complication of graft necrosis occurred in our case. We suggest that, in large defects, reconstruction with vascularized grafts is a safer method than free grafts. Using a reconstruction plate can maintain the facial contour in cases where the graft procedure failed at the postoperative period.

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