Resection of an ameloblastoma in a pediatric patient and immediate reconstruction using a combination of tissue engineering and costochondral rib graft

A case report

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Ameloblastoma is an odontogenic tumor predominantly occurring in patients who are in their 20s and 30s.[1] It is estimated that approximately 10% to 15% of ameloblastomas occur in patients who are younger than age 18 years.[1] Resection of larger tumors can leave patients with continuity defects, poor oral functioning, and facial deformities. Autogenous cancellous bone marrow grafts, distraction osteogenesis, or free flaps are used for immediate reconstruction; however, each technique has its morbidity and downfalls.[2] Surgeons have reported using a combination of osteoconductive (allogeneic bone), osteoinductive (recombinant human morphogenetic protein-2 [rhBMP-2]), and osteogenic (bone marrow aspirate) agents to reconstruct large benign tumor defects in adults.[3,4] In this article, we describe a pediatric patient who underwent resection of a mandibular unicystic ameloblastoma with immediate reconstruction using a costochondral rib graft, allogeneic bone, bone marrow aspirate concentrate (BMAC), and rhBMP-2.

CASE REPORT

With the approval of the institutional review board of the University of Texas Health Sciences Center at Houston, Houston, Texas, we report a case of a healthy 11-year-old girl who was referred to our clinic for evaluation of a unilocular radiolucent lesion of the posterior left mandible.

ABSTRACT

Background and Overview. Ameloblastoma is an odontogenic tumor predominantly occurring in patients who are in their 20s and 30s. Approximately 10% to 15% of ameloblastomas occur in patients younger than 18 years. Although it is a benign tumor, an ameloblastoma can have a devastating effect on children both physically and emotionally. The aim of this case report is to demonstrate how tissue engineering and surgical techniques can minimize morbidity and recovery time after extirpation and immediate reconstruction of a mandibular ameloblastoma.

Case Description. An 11-year-old girl was referred for surgical evaluation of a lesion found on a routine dental radiograph. Resection of a mandibular unicystic ameloblastoma resulted, including immediate reconstruction using a costochondral rib graft, allogeneic bone, bone marrow aspirate concentrate, and recombinant human morphogenetic protein-2. One year postoperatively, the patient had no evidence of recurrence as well as excellent mandibular bone height and width with good facial form. The patient has returned to her daily life without any disabilities or disfigurement.

Conclusions and Practical Implications. Dentists are typically the first health care providers to discover oral pathology in patients. The coordination of care by the dental care providers and the oral and maxillofacial specialist was key to the successful outcome for this patient. With biotechnology and surgical techniques, the dental surgeon can extirpate an ameloblastoma and reconstruct the mandible defect to the ideal shape and size with minimal morbidity and recovery time.

Key Words. Tissue engineering; oral and maxillofacial surgery; oral and maxillofacial pathology; neoplasms; pediatric dentistry; oral surgical procedures; bone grafting; bone marrow transplantation; bone substitutes.

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Figure 1. Orthopantomograph of a patient with a tooth displaced toward the angle of the mandible, extensive bony resorption, and lingual displacement of teeth nos. 18 and 19.

Figure 2. Costochondral rib graft secured to custom reconstruction plate before insertion.

Figure 3. Resected ameloblastoma from the left mandible.

The lesion had been found on a routine dental radiograph. We obtained a cone-beam computed tomographic scan, which revealed a 3.4 × 4.2 × 3.1-centimeter, well-defined, osteolytic lesion associated with an impacted tooth no. 17. The tooth was displaced toward the angle of the mandible with extensive bony resorption and lingual displacement of teeth nos. 18 and 19. The patient also had marked facial asymmetry and vestibular expansion of the alveolus (Figure 1); however, neither erythema, ulceration, nor neurologic deficits were noted. We performed an endoscopically assisted incisional biopsy while the patient was under general anesthesia, which yielded the histologic diagnosis of ameloblastoma. We created a treatment plan for surgical resection with immediate reconstruction.

Due to the size of the anticipated defect and desire for immediate reconstruction, we consulted with the patient and her family for their consent to the off-label use of rhBMP-2 and BMAC. We explained all other options in full, and we made the decision to proceed. We harvested a costochondral rib graft to reconstruct the mandibular condyle (Figure 2), and we constructed the 12-cm continuity defect (Figure 3) with

- a resorbable mesh (poly-[D,L]-lactide co-polymer) (KLS Martin USA);
- 120 milliliters of allogeneic corticocancellous bone (Musculoskeletal Transplant Foundation);
- 12 milligrams of bone morphogenic protein (BMP) (Medtronic);
- 100 mL of BMAC.

The final pathology report revealed unicystic ameloblastoma with focal intraluminal plexiform features. The patient did well postoperatively and remained in maxillomandibular fixation for 3 weeks. She spent a total of 2 nights in the pediatrics unit. One year postoperatively, we provided the patient with an Essix retainer to act as a removable space maintainer while she awaits a removable partial denture in the future. A cone-beam computed tomographic scan at the 1-year follow-up examination showed adequate bone with excellent height, width, and arch coordination for endosseous dental implant placement (Figure 4).

The treatment planned is for the patient to receive dental implants on reaching skeletal maturity. At the same time of the implant surgery, the patient also will have some mandibular recontouring due to excessive bone formation (Figures 5 and 6).

DISCUSSION

Multiple treatment modalities for ameloblastoma are discussed in the literature. Common treatment options include enucleation, curettage, enucleation with chemotherapeutics (Carony's solution), and segmental or marginal resection.[5-7] Though some sporadic reports in the literature involve marsupialization, this treatment modality alone is not intended for treatment of locally aggressive tumors such as ameloblastoma.[6] A review of the literature found a significant recurrence rate in the

ABBREVIATION KEY. **BMAC:** Bone marrow aspirate concentrate. **BMP:** Bone morphogenic protein. **rhBMP-2:** Recombinant human morphogenetic protein-2.