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CASE REPORT

Pediatric/Craniofacial

Diagnosis and Treatment of a Benign Pediatric Mandible Tumor

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Summary: Pediatric mandible tumors are rare and generally benign but can be locally aggressive. Diagnosis and treatment involve tumor identification, excision, and subsequent reconstruction. Successful reconstruction should address form and function of the mandible while minimizing morbidity. The authors review the diagnosis and treatment of a benign pediatric mandible tumor and the use of nonvascularized rib graft for mandible reconstruction. The subject of interest is an 8-year-old boy who presented with progressive left-sided facial swelling and examination findings concerning for a mandibular neoplasm. A large bony tumor of the left mandibular ramus and condyle was identified on computed tomography scan. Needle aspiration was performed but was nondiagnostic. The patient underwent en bloc resection and immediate reconstruction utilizing nonvascularized rib graft. Pathologic analysis demonstrated a benign fibro-osseous lesion consistent with fibrous dysplasia, ossifying fibroma, or aneurysmal bone cyst. The patient had excellent recovery of jaw function and resumption of solid diet 5 weeks after reconstruction. Mandibular defects >6 cm in length often require free vascularized bone flaps for reconstruction; however, these procedures can have greater morbidity in the skeletally immature patient. Nonvascularized rib graft is a viable alternative that also allows for reconstruction of the mandibular condyle using the costocartilagenous cap of the harvested rib. The authors present this case as an example of a rare pediatric head and neck tumor and review of the approach to diagnosis and treatment, including special considerations for complex pediatric mandibular reconstruction. *(Plast Reconstr Surg Glob Open 2019;7:e2452; doi: 10.1097/GOX.0000000000002452; Published online 15 October 2019.)*

Pediatric mandibular tumors are rare and generally benign but can be locally aggressive. They are most common during the second decade of life, often presenting with pain and facial deformity. Diagnosis relies on clinical suspicion, careful physical examination, and advanced imaging. In cases of aggressive tumors causing significant pain or deformity, treatment typically requires en bloc tumor excision.[1,2] Resulting defects vary from minimal bony loss to segmental mandibular defects. Restoration of bony continuity is recommended to optimize mastication, speech, and maintain symmetry of the lower third of the face.[3] The authors present a rare and interesting case of a pediatric mandible tumor, and review the approach to diagnosis and treatment.

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CASE PRESENTATION

An 8-year-old boy presented to the emergency department for evaluation of left-sided facial swelling. He was diagnosed by history and examination alone with viral parotitis and treated at home with nonsteroidal anti-inflammatory medications and sialagogues. No imaging studies were performed initially. When his symptoms failed to improve, he was referred to the otolaryngology clinic where examination findings were concerning for a mass involving the left mandibular ramus. A computed tomography scan revealed a large, multilocular, expansile lesion replacing the left mandibular ramus and condyle (Fig. 1).

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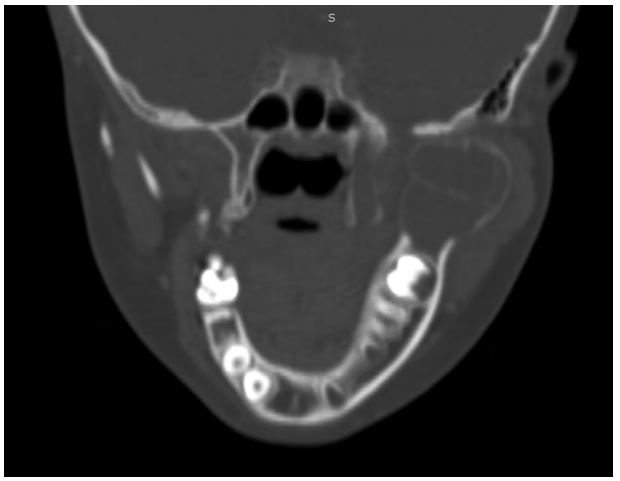

Fig. 1. Coronal CT scan with contrast demonstrating a multilocular lesion of the left mandibular ramus and condyle. CT indicates computed tomography.

Needle aspiration of the tumor returned only bloody fluid which was nondiagnostic on pathologic analysis. Given that the appearance of the lesion on computed tomography imaging was suggestive of a benign tumor, the decision was made to proceed with resection and bony reconstruction. Before resection, angiography of the lesion was performed but a target vessel for embolization was not identified. Subsequently, the patient underwent segmental mandibulectomy via a transcutaneous approach. Intraoperative frozen section analysis demonstrated no evidence of malignancy. The resulting defect was reconstructed using autologous nonvascularized rib graft. The costocartilagenous cap of the harvested rib was utilized for condylar reconstruction. Figure 2 shows the resected specimen. Initial histopathologic analysis did not identify a definitive diagnosis, so specimens were sent to a second institution for further review. Histopathologic analysis at that institution returned a differential diagnosis of fibrous dysplasia, ossifying fibroma, and aneurysmal bone cyst. On closer review with the pathologist assigned to the case, it was determined that the specimen demonstrated areas of blood islands, reactive granulation, and multinucleated giant cells as shown in Figure 3, which is most consistent with an aneurysmal bone cyst.

Postoperatively, dental occlusion was maintained with rigid maxillomandibular fixation for 1 week followed by elastic physiotherapy for an additional 4 weeks. The patient was started on a liquid diet on postoperative day 1 and was able to maintain adequate nutrition without other feeding access. At 5 weeks, panoramic radiographs demonstrated stability of the rib graft construct with articulation of the neocondyle in the glenoid fossa and the patient was advanced to a soft solid food diet. Three months after surgery, he resumed a regular diet without restrictions. At approximately 1 year after surgery, the patient reported no pain or malocclusion and panoramic radiographs showed stability and bony remodeling of the rib graft construct with articulation of the neocondyle in the glenoid fossa (Fig. 4).

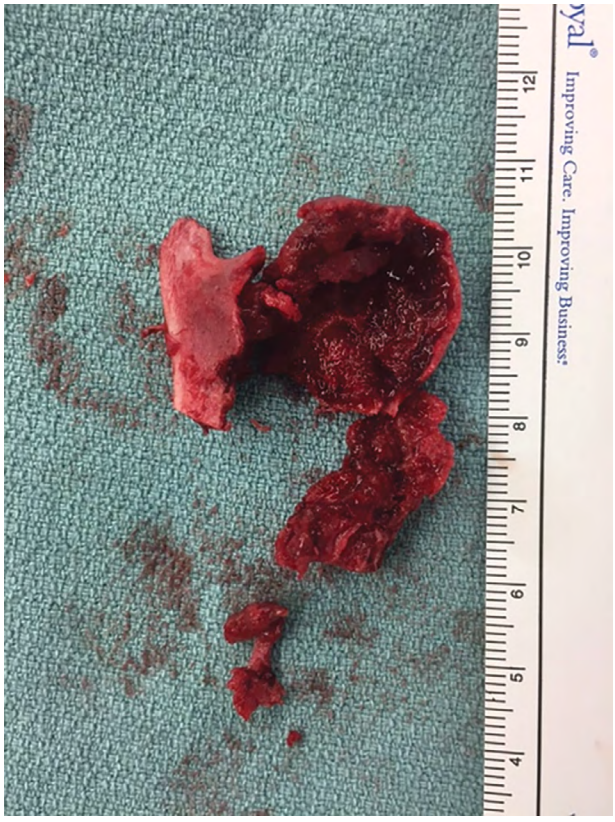

Fig. 2. Photograph of resected tumor.

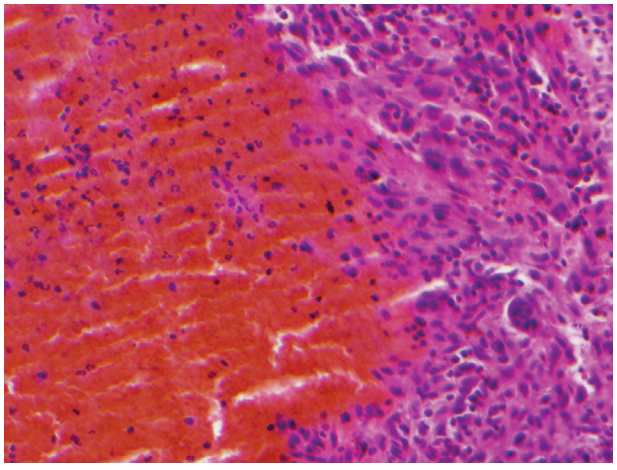

Fig. 3. Hematoxylin and eosin stain 200× photomicrograph of specimen with blood island, granulation tissue, and multinucleated osteoclast. H&E indicates hematoxylin and eosin.

DISCUSSION

As with all rare conditions, the initial diagnosis of a pediatric mandible tumor can be challenging.[1] Following discovery of such a tumor, a biopsy should be performed to obtain tissue for pathologic analysis, as the approach to treatment may vary depending on the type of tumor and whether the lesion is benign or malignant.[2] Although needle aspiration of the tumor in this case did not provide a definitive diagnosis, the fact that blood was aspirated from

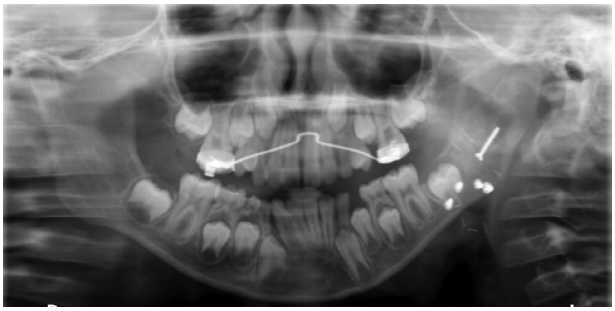

Fig. 4. Panoramic radiograph of reconstructed mandible at 1 y.

the tumor cavity was suggestive of the diagnosis of aneurysmal bone cyst as these lesions are often blood-filled.

In the case of a rapidly enlarging aneurysmal bone cyst, en bloc resection followed by reconstruction is required.[1] Although reconstruction plates or screws are often utilized for bony stabilization of autologous bone grafts or flaps, the use of alloplastic materials alone is generally discouraged as they do not allow for facial growth and are prone to hardware loosening and extrusion.[4,5] The size and location of a segmental mandibular defect determine the type of bony reconstruction needed, with larger defects requiring cortical bone using either a cortical bone graft or osteocutaenous free flap spanning the defect for structural support, and in this case for temporomandibular joint (TMJ) reconstruction.

Here the authors elected to use nonvascularized rib grafts for mandible and TMJ reconstruction. Several studies have demonstrated durability and functional improvement after utilizing nonvascularized rib grafting for mandible reconstruction in both adult and pediatric patients.[2,6–8] It should be noted, however, that rib graft growth has been shown to be unpredictable and that undergrowth and resorption and bony overgrowth have been reported.[8,9] Other potential benefits of this technique include decreased donor site morbidity, operative time, and hospital length of stay compared with microvascular reconstruction techniques.[6,9,10] The main disadvantage of nonvascularized rib graft for mandible reconstruction is the limited bone stock provided. However, the segment of mandible being reconstructed in this case was non–tooth bearing such that bone height for future dental implants was not a concern. In addition, a strut configuration was utilized to increase the strength of the reconstruction.

CONCLUSIONS

This case provides an example of the approach to the diagnosis and treatment of a benign but locally aggressive pediatric mandible tumor. Accurate diagnosis with advanced imaging and tissue biopsy should guide the surgeon's plans for treatment, including tumor excision and reconstruction. In the pediatric patient, it is particularly important to not only restore form and function of the growing mandible but also limit any donor site morbidity during reconstruction. Although larger defects or those involving bone and soft tissue may require free flap reconstruction, nonvascularized rib grafting is a well-described alternative for moderate size defects of the mandible and can produce an aesthetically pleasing and stable result. The costochondral graft is particularly well suited for TMJ reconstruction providing cartilage for an articular surface and the potential for compensatory growth.

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