Reconstruction of Ramus-Condyle Unit with Two Rib Grafts: Report of a Case

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Introduction: Ramus-condyle unit (RCU) reconstruction is one of most challenging procedures in maxillofacial surgery. Autologous graft, custom made prosthesis and transport disc distraction are some treatment options. **Case Report:** In this study, we reported a mandibular DF in a 9-year-old child with condylar involvement. Our treatment plan was segmental resection and reconstructed RCU with 2 rib grafts. **Results:** After one year of follow-up, there is no evidence of recurrence. Mandible has symmetrical appearance and normal centric and eccentric movement. **Conclusion:** Rib graft is recommended for RCU reconstruction, especially in children in the growing stage.

Keywords: Desmoplastic Fibroma; Costochondral Graft; Reconstruction; Hemimandibulectomy; Ramus-condyle Unit

Introduction

Desmoplastic fibroma (DF) is a rare benign non-odontogenic tumor and locally aggressive tumor with the origin of connective tissue in bones, which occurs in children in the first two decades of life (1,2). Majority of DF has been found in long bones. In maxillofacial region, mandible is the most common site of involvement (3). The clinical manifestation of DF is slow growing and painless swelling of the affected site. Radiographically, DF is a unilocular or multilocular radiolucent lesion with frequently cortical perforation. The clinical and radiographical features of DF is not specific and histological evaluation is needed for definitive diagnosis (4). Because of the aggressive nature of this lesion, complete resection is recommended. Reconstruction of RCU defects has always been challenging especially in children due to the growth stage. Some treatment options are costochondral graft, sternoclavicular graft, total joint custom-made prosthesis and transport disc distraction. The goal of reconstruction is facial symmetry, removing malocclusion and normal mandibular movement. These techniques have some advantages and disadvantages. For example, autologous graft has growth potentials despite inducing donor site morbidity. One of disadvantage of Transport disc distraction is the poor control of vector (5,6). In this study, we presented a case of mandibular DF with condylar involvement in a child that reconstructed RCU with 2 rib grafts after resection.

Case Report

A 9-year-old boy was referred to Taleghani Hospital maxillofacial clinic with a history of progressive asymmetric painless growth at the left mandibular angle and ramus. In the past 6 months, the swelling was progressive in size and firm in consistency. The patient's general examination revealed no pathological alteration of any other body structure. The patient presented good dental health, with an absence of decay, periodontal disease or oral mucosa alteration. He complained of mild pain over the swelling, but had no difficulty opening the mouth.

Clinical extra-oral examination revealed firm swelling on the left side of the jaw with well-defined border, and fixed to the mandible. Skin over the swelling was normal with no alteration in sensation in the area.

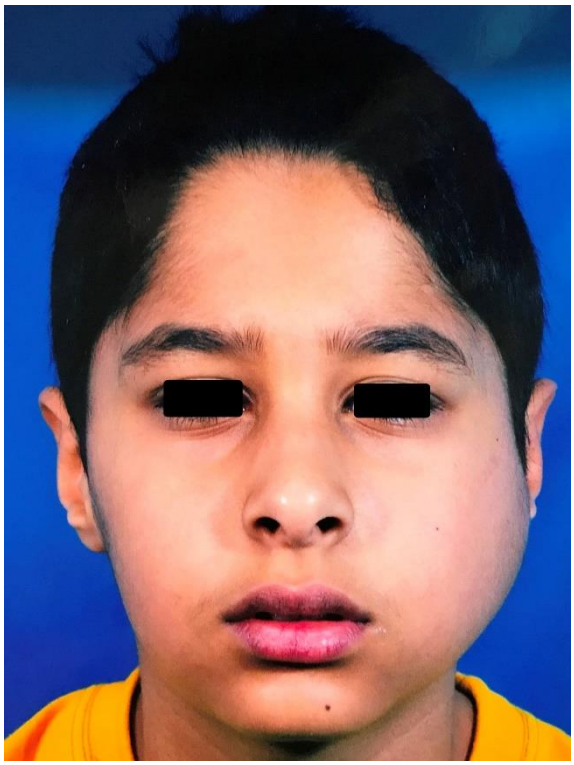

Figure 1. Extra oral appearance of lesion in left side of mandible

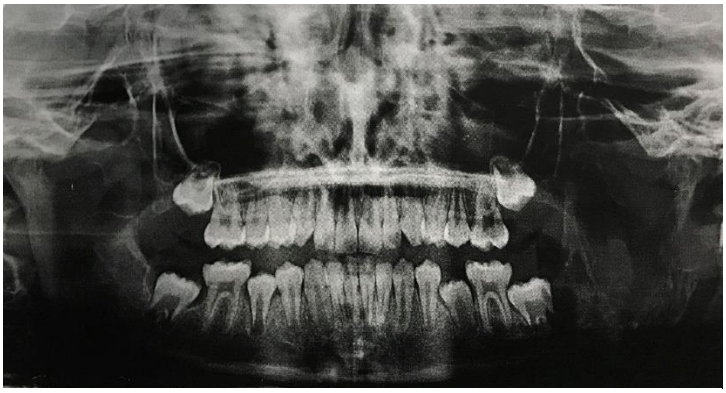

Figure 2. Panoramic view of patient

Intra-oral examination revealed a solitary bony hard swelling with obliteration of the left buccal vestibule in relation (Figure1). The panoramic radiograph showed an multilocular radiolucency in the left mandibular angle, ramus and condyle area (Figure 2). A computerized tomography (CT) scan demonstrated buccal and lingual cortical plate expansion, and the area demonstrated a 6-cm lesion at the l eft ascending ramus, extending from the angle of the left mandible to the sigmoid notch, with evidence of buccal and lingual cortical perforation and soft tissue invasion (Figure 3).

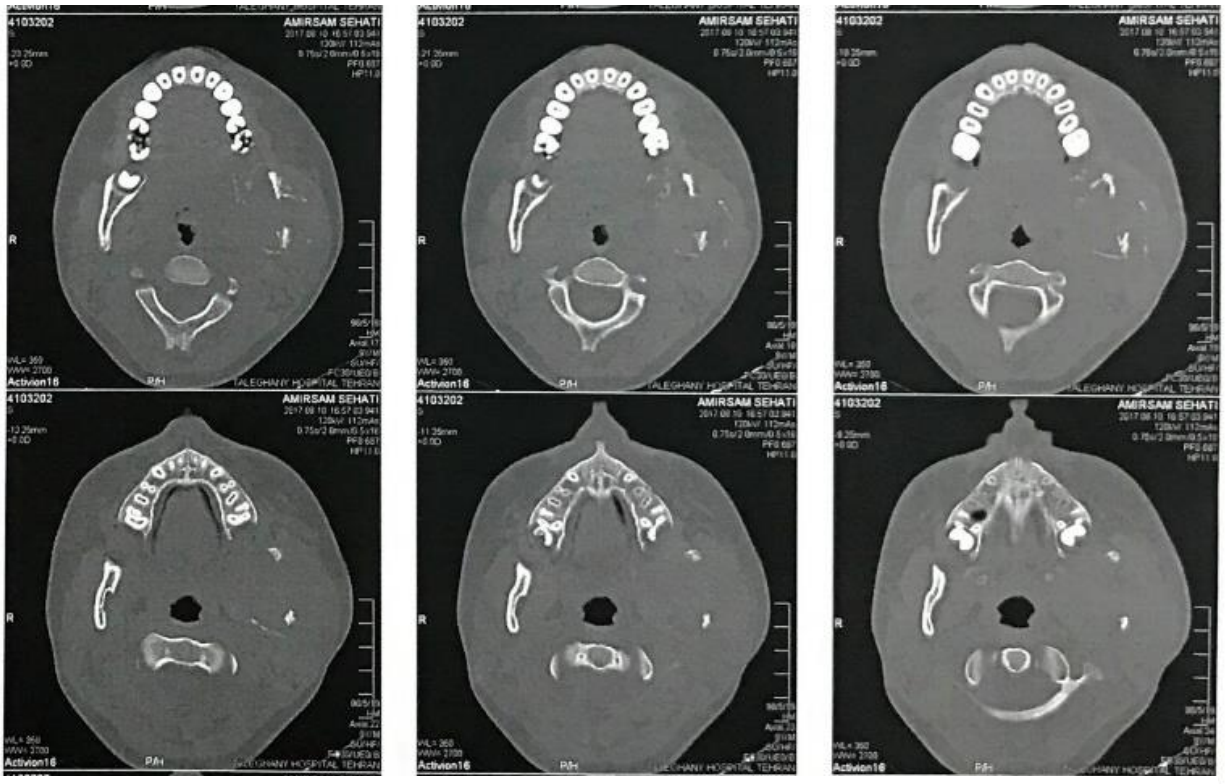

Figure 3. Computed tomography (CT) of patient

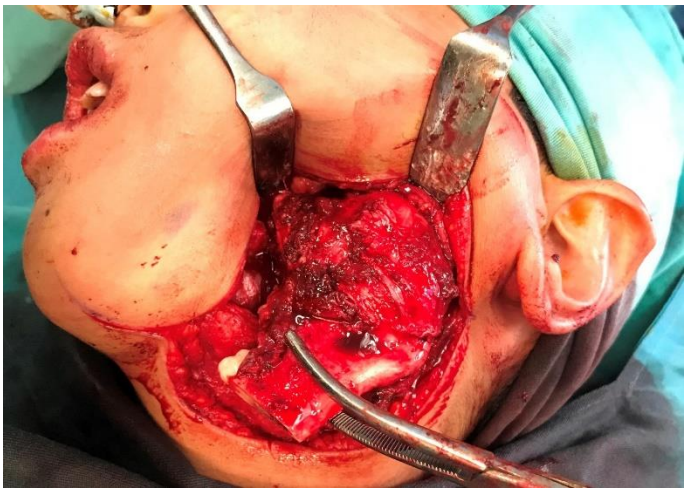

Figure 4. Segmental resection of lesion from extra oral approach

The initial histopathological diagnosis of a DF was obtained by an incisional biopsy. The subsequent surgical removal of the lesion was performed through a submandibular approach and segmental resection (hemimadibulectomy) from the mesial of the first molar with 1 cm margin was done, and removing the angle, condyle/ramus unit with a supraperiosteal cuff around the perforation site was done (Figure 4). The area was immediately reconstructed with costochondral grafts from the right 6th for the reconstruction of condylar unit area, and costobone graft 7th ribs for reconstruction of part of body and angle area. These two rib grafts were secured with two mini plates and one lag screw. In the angle are we overlapping a two segment of rib bone graft for better reconstruction of angle area (Figure5).

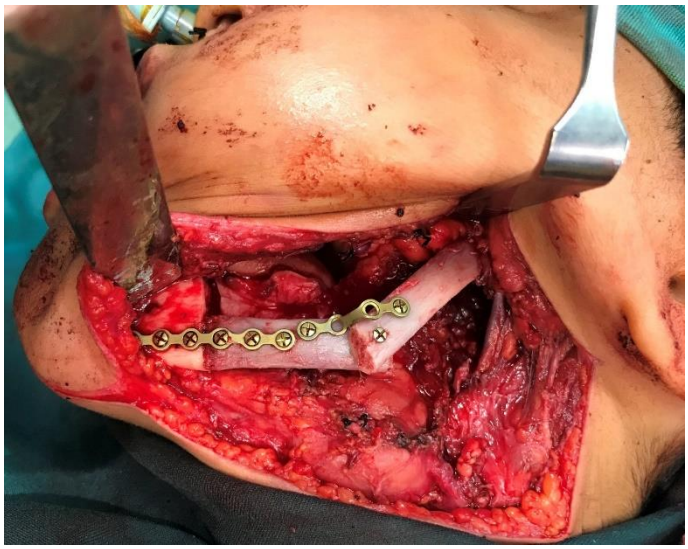

Figure 5. Two rib grafts were used for reconstruction of defect

Risdon wire was performed on the remaining mandibular teeth and maxillary teeth for elastic therapy. Physiotherapy was started 1 week after the surgery to achieve normal mouth opening and extrusive & laterusive mandibular movement.

Results

In the final pathology, we noted a desmoplastic fibroma with negative margins (Figure 6).

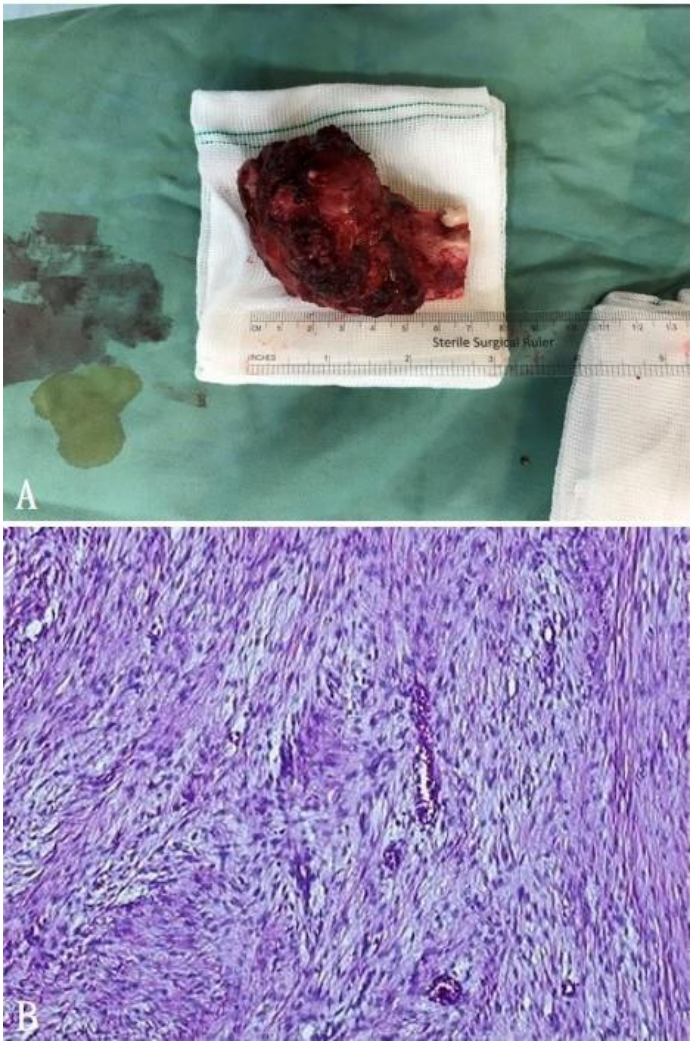

Figure 6. (A) Resected section of mandible with lesion; (B); Histologic view of lesion

The patient had an uneventful postoperative course. There were no signs of infection of malunion. The patient was visited every 2 weeks for the first 2 months and then monthly for 4 months

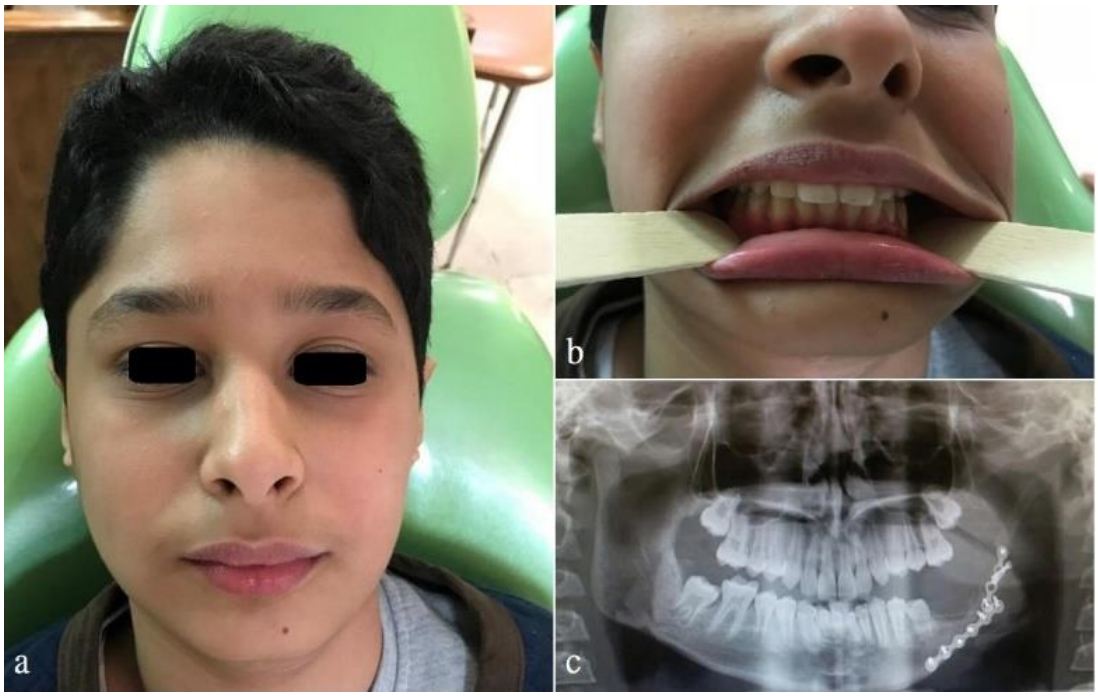

Figure 7. (A) patient appearance reveals symmetrical growth of costochondral graft and there is no asymmetry after 1 year follow up; (B) Normal occlusion after 1 year follow up; (C) Panoramic view of patient after one year

and every 3 months until 1 year postoperatively. After one year, he was noted to have normal mandibular movements (maximum mouth opening, extrusive and laterusive movement), normal occlusion, symmetric growth, excellent bony union with no evidence of recurrence (Figure 7).

Discussion

Desmoplastic fibroma is a rare tumor in maxillofacial region, representing less than 1% of all bone tumors (2). More than 75% of this lesion was seen in before 30 years of age (7).

Histological evaluation is crucial to characterize DF. Microscopically, DF is composed of spindled and elongated fibroblasts separated with densely collagenous fibers. Cellularity may vary from low to moderate. We found no bone formation, cellular atypia, or mitotic figures (2,8), hence differentiating DF from malignant lesions. Because of the presence of spindled cells, DF was placed in differential diagnosis with other spindle-cell lesions, such as non-ossifying fibroma, odontogenic fibroma, fibrous dysplasia and low-grade fibrosarcoma. Some authors reported that some IHC markers such as vimentin, α-smooth muscle actin (certain cells), S-100, Ki-67, and factor XIIIa could be positive in DF (8,9). For example, one study shows that S-100 is usually negative for DF (9).

Radiological findings of DF are nonspecific. DF is a radiolucent lesion with well-defined margins and unilocular and multilocular appearance (usually "soap bubbles" appearance) (3,4,10). Cortical perforation may be presented. These radiological appearances were found in other lesions such as odontogenic tumors (ameloblastoma, myxoma), aneurismal bone cyst, central hemangioma, non-ossifying fibroma and chondromyxoid fibroma. CT is recommended for the evaluation of lesion extension and cortical perforation (11). Most of the DF was found in the posterior of mandible, but the condylar region is an uncommon site. Our case was a 9-year-old boy with DF in the posterior of mandible and condylar involvement. Due to a high recurrence rate for DF, aggressive treatment is better than the conservative surgery. Most studies suggest complete wide resection with free margins to minimize the chance of recurrence especially in huge lesions (2,12,13). Enucleation & curettage is not recommended due to high recurrence rate (approximately 40%) (3). Conservative treatment can be used only in small lesions without cortical destruction. Reconstruction of defect after resection is a major step in any treatment. In mandibular resection, providing continuity of mandible, symmetry and normal pre-surgical occlusion is

crucial. Because DF is more frequent in children, the growth potential of patients should be considered for reconstruction.

In our case, we used costochondral rib graft for the reconstruction of RCU, since in this patient condyle was a growth center for mandibular development. Related literature showed that costochondral graft is one of the best options for condylar reconstruction in the growing patient due to its cartilagenous portion that can remodel and adapt as a condyle. However, donor site morbidity and unpredictable growth pattern are some disadvantages of costochondral graft1 (4,15). Some studies recommended the use of free microvascular flap for reconstruction. Growth potential of free flap is questionable. According to investigations, free flap does not have a growth potential, while some authors believed that free flap can growth with mandibular development in animal study (16). For the reconstruction of RCU, in literature, there are some treatment options. Some investigations used transport disc distraction for reconstruction. Mehrotra et al., (17) reported 8 cases of RCU reconstruction with transport distraction. They noted acceptable mouth opening and minimal pain. Two studies compared autologous bone graft and transport distraction in RCU reconstruction (5,6). Kaur et al., (5) concluded that both techniques have acceptable results, but Kohli et al., (6) found that bone grafts are superior to transport distraction. The advantage of our technique for RCU reconstruction is there is no other donor site and second ribs graft harvested from incision that used for costochondral graft harvesting. After a 12-month follow-up, there is no evidence of recurrence, and mandibular opening and eccentric movement are normal. Mandible has symmetrical appearance and there is no overgrowth or undergrowth in the costochondral graft.

Conclusion

As a conclusion, rib grafts as a costochondral graft and the autogenous graft are recommended for the reconstruction of RCU defects especially in children.

Conflict of Interest: 'None declared'.

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