Free fibula flap for management of juvenile ossifying fibroma: A case report

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Abstract

Juvenile ossifying fibroma is a benign fibro-osseous neoplasm seen usually below 15 years of age. It is an aggressive expansile lesion that requires radical resection to prevent recurrences. The acquired defects if untreated can lead to facial asymmetry and functional difficulties thus require reconstruction with an aim to achieve better esthetics and functional outcome in a growing child. A 9-year-old male patient reported with 3 cm × 4 cm sized hard swelling on the left body of the mandible. On the radiological examination, tumor showed well-defined radiopaque-radiolucent lesion from the distal aspect of the developing 13 to the mandibular angle, ramus region on the left side. After the clinical, radiological and histologic analysis, it was diagnosed with juvenile ossifying fibroma. The patient underwent segmental mandibulectomy and reconstruction with free fibula flap. A conservative approach in a tumor with a proven high recurrence is unintentional planning of second surgery. Ablative surgery in pediatric age has to be planned meticulously as they impact the development of child. Free flaps with epiphyseal growth centers have the potential for uninterrupted growth of the jaw.

Keywords: Fibula free flap, juvenile ossifying fibroma, ossifying fibroma

Introduction

Juvenile ossifying fibroma is a benign fibro-osseous neoplasm seen in children below 15 years of age. It characteristically presents as a clinically asymptomatic aggressive expansile lesion. It has a greater predisposition to maxilla than mandible. The radical resection is associated with the least recurrence rates. The aggressive nature of the tumor requires radical treatment with the reconstruction of the jaw. If these acquired defects are untreated, they can lead to facial asymmetry and functional difficulties. Reconstruction along with rehabilitation provides better esthetic and functional outcomes relieving the growing child of the psychological, physical trauma of radical resection of the jaw. The options are reconstruction plates and free flaps. Reconstruction plates provide good anchorage and excellent biocompatibility. They are frequently associated with post-operative complications such as plate exposure, plate fracture, dehiscence, need for second surgery, and infection. Free flaps have an advantage of better bone stock and ability to design skin paddle. The selection of donor sites in a child is based on the presence of epiphyseal growth centers and long-term development unlike adults were comorbidities play a central role.

Case Report

A 9-year-old male patient reported to us with a chief complaint of pain and swelling over the left side of the lower jaw for 6 months. On examination, there was gross facial asymmetry noted in the lower third of the face with a well-defined solitary swelling measuring approximately 3 cm × 4 cm extending anterio-posterior from commissure of the lip to the angle of mandible and superio-inferiorly extending from line joining corner of the mouth to tragus of the ear to about 1 cm inferior to the lower border of mandible [Figure 1]. Overlying skin appeared normal. On palpation, the swelling was tender and bony hard in consistency with well-defined borders. Intra-orally vestibular obliteration was noted from the left primary second molar to permanent first molar region buccally and lingually [Figure 2]. Egg shell crackling was elicited on palpation of the buccal cortical plate. The left single submandibular lymph node was palpable, non-tender, soft, and mobile on the left side.

Orthopantomogram revealed a predominantly well-defined radiopaque-radiolucent lesion extending from the distal aspect of the developing 13 to the mandibular angle, ramus region on the left side. Superiorly it was extending from the alveolar
bone to the inferior border of the mandible. The borders were well-defined with no pathological root resorption, there was the expansion of the lower border of mandible with a thin rim of sclerotic margin in the angle region.

Multidetector computed tomography face (plain and contrast) revealed a fairly well-defined large expansile lytic lesion with thin internal septations involving the body of mandible on the left side, approximately measuring $3.0 \times 2.7 \times 3.4$ in cm, contrast study showed intralesional soft-tissue components [Figure 3]. Routine blood investigations were done which was within normal limits. An incisional biopsy was performed and was reported as juvenile ossifying fibroma.

The patient underwent tracheostomy. Apron incision, extending from symphysis menti to mastoid region, was used. The bony lesion was solid expansile, well defined with areas of erosion of cortical bone [Figure 4]. Segmental mandibulectomy sparing the ramus of the left mandible was performed. The affected part of the jaw was $7 \text{ cm} \times 2 \text{ cm}$ and the defect was reconstructed using free fibula flap, first of its kind in India [Figure 5]. A vascularized fibula flap with a skin paddle was harvested, and the vascular pedicle was anastomosed end to end to the facial artery and end to side to the external jugular vein. The skin paddle was inset extra

Figure 1: Extra oral facial deformity

Figure 2: Intraoral vestibular obliteration

Figure 3: Mandibular cortical expansion on computed tomography with contrast

Figure 4: Intraoperative surgical resection

Figure 5: Free fibula flap reconstruction
orally, and the viability of the fibula flap was followed by monitoring the attached skin paddle.

Discussion

The term juvenile ossifying fibroma was first described by Jhonson in 1952.\(^3\) JOF arises from periodontal ligament. Midface involvement is common (90%) and mandible is involved in 10% of cases.\(^4\) Radiographically, it presents as a well-defined multilocular or unilocular radiolucent lesion with occasional radiopacity. The various modalities of treatment are enucleation, curettage, and radical resection. The tumor has a good prognosis in adults but in children, it is associated with high recurrence. The recurrence rate of curettage was 71.4% and 50% for local excision was high. The recurrence rate for resection is 4%.\(^5\) The high recurrence has been attributed to the aggressive nature, tumor residue after curettage, growth, and developmental potential of the child.

Although the recurrence rate was low for resection, it is morbid and affects the quality of life of the child. One of the major concerns about resection is the development of jaw in children. The craniofacial development is dependent on the relationship between the maxilla and mandible and osseous defects of the jaw can hamper the growth.

According to Chuong and Kaban,\(^6\) children who undergo resection of the jaw and reconstruction with an autogenous bone appear to grow normally.

Hence, we planned to reconstruct the mandible with a free fibula flap. Fibula flap has the ability to grow with the native mandible and thus dental rehabilitation is possible. The growth of the fibula and mandible is age-dependent. Epiphyseal plates are growth centers in mandible and fibula. The fusion of epiphyseal plates occurs between 15 years for boys and 18 years for girls. An anthropological study observed, width and height of bone increases between 1 and 5 years, whereas depth increases only after 5 years. Later between 8 and 12 years, there is a rapid increase in width, height, and depth.\(^7\)

There are certain concerns such as vessel caliber and vasospasm in pediatric fibula free flap which have not been established. In our case, the caliber of the pedicle was appropriate and flap uptake was satisfactory. Long-term donor site morbidity is low.\(^8\)

Conclusion

A conservative approach in a tumor with a proven high recurrence is unintentional planning of second surgery. Ablative surgery in pediatric age has to be planned meticulously as they impact the development of the child. Free flaps with epiphyseal growth centers have the potential for uninterrupted growth of the jaw.

Clinical Significance

Juvenile ossifying fibroma is categorized as a highly aggressive tumor rarely seen in the mandible with a common line of treatment being enucleation/curettage. This treatment is not satisfactory in a growing child and would result in recurrence; hence, a radical resection and reconstruction with a fibula free flap would be appropriate as it would take care of the developing jaw with minimal donor site morbidity.

References

1. Seol GJ, Jeon EG, Lee JS, Choi SY, Kim JW, Kwon TG, et al. Reconstruction plates used in the surgery for mandibular discontinuity defect. J Korean Assoc Oral Maxillofac Surg 2014;40:266-71.
2. Genden EM, Buchbinder D, Chaplin JM, Lueg E, Funk GF, Urken ML. Reconstruction of the pediatric maxilla and mandible. Arch Otolaryngol Head Neck Surg 2000;126:293-300.
3. Jhonson LC. Bone pathology seminar. Am Acad Oral Pathol 1952;18:13-7.
4. Johnson LC, Yousefi M, Vinh TN, Heffner DK, Hyams VJ, Hartman KS. Juvenile active ossifying fibroma. Its nature, dynamics and origin. Acta Otolaryngol Suppl 1991;488:1-40.
5. Liu Y, Shan XF, Guo XS, Xie S, Cai ZG. Clinicopathological characteristics and prognosis of ossifying fibroma in the jaws of children: A retrospective study. J Cancer 2017;8:3592-7.
6. Chuong R, Kaban LB. Diagnosis and treatment of jaw tumors in children. J Oral Maxillofac Surg 1985;43:323-32.
7. Farkas LG, Posnick JC, Hreczko TM. Growth patterns of the face: A morphometric study. Cleft Palate Craniofac J 1992;29:308-15.
8. Barla M, Polirsztok E, Peltié E, Jouve JL, Legré R, Dautel G, et al. Free vascularised fibular flap harvesting in children: An analysis of donor-site morbidity. Orthop Traumatol Surg Res 2017;103:1109-13.