CASE REPORT

Reconstruction using free fibular flap in surgical treatment of maxillary juvenile ossifying fibroma in children: A case report

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SUMMARY

Fibro-osseous lesions are rare benign tumours, with ossifying fibroma (OF) being the most common type in the head and neck region. Fibula free flap is the preferred choice for maxillo-mandibular defect reconstruction in paediatric patients, as it provides both bone and soft tissue components. We report a case of a 10-year-old girl presenting with a lump on the right cheek for the past six months. Surgical management of OF depends on clinical and radiological manifestations and typically involves enucleation, curettage, or resection. The aggressive nature and high recurrence rate make radical surgery the preferred therapy. Defects in the maxilla resulting from tumour removal can lead to serious functional and aesthetic deformities. Reconstruction using a free fibula flap in children with OF yields satisfying results with minimal postoperative morbidity while maintaining aesthetics and functionality over time. A multidisciplinary team is required in managing such complex cases to ensure a good outcome.

INTRODUCTION

Ossifying fibroma (OF) is the most common benign fibro-osseous lesion of the head and neck region, commonly affecting patients aged 20 to 40, though it is sometimes also found in children (juvenile ossifying fibroma/JOF). JOF is divided into trabecular type (juvenile trabecular ossifying fibroma/JTOF) and psammomatoid type (juvenile psammomatoid ossifying fibroma/JPOF). JTOF more frequently affects the maxilla and shows erosion and invasion into the surrounding structures, while JPOF mostly affects the sinonasal.[1]

Patients with OF usually present with a painless, slow-growing mass but might also be accompanied by pain or paraesthesia, face deformity or asymmetry, proptosis, sinus obstruction, teeth displacement, or intracranial complications. On the other hand, JOF tends to be painless while growing rapidly and causes destruction to adjacent structures.[1,2] Surgical management involves enucleation, curettage, or resection. Small lesions are treated conservatively with enucleation or curettage, while larger lesions require radical surgery and reconstruction. Mandibular OF are typically well-defined and can be surgically removed with ease, while maxillary OF are more challenging to treat due to possible expansion into the maxillary sinus.[1] Reconstructive surgery of the maxilla might be required to restore functionality and aesthetic following tumour removal, ensuring normal functions of the stomatognathic system, separating oral from sinonasal cavity, and restoring normal facial contours. Reconstruction using a fibula flap is an effective method of treating maxillo-mandibular defects in adult patients and is reported to have a high recovery rate with low postoperative morbidity in paediatric patients. Complications at 30 and 90 days following surgery were reported in less than 20% of patients, the most common being minor wound healing issues, such as cellulitis and wound dehiscence. The harvesting of fibula flaps does not cause any long-term orthopaedic complications, as previous studies reported paediatric patients with fibula flaps to have normal limb length and gait.[2] Considering the complexity of such cases, the involvement of a multidisciplinary team is required to ensure a good outcome. The objective of this study is to evaluate the benefits of fibula free flap in maxilla-mandibular reconstruction in children with oncological issues. The study aims to highlight the postoperative outcomes of the fibula-free flap, focusing on functional and aesthetic aspects.

CASE PRESENTATION

A 10-year-old girl complained of a growing lump on her left cheek for the past six months. The lump originated in her left upper jaw, on the gum of her first molar, after a tooth extraction. At the first presentation at the hospital, the lump was approximately the size of a baseball and was painless, but after a month grew into the size of a basketball, covering her oral cavity, causing difficulties in feeding and significant pain due to the mass pressing into surrounding structures. On physical examination, a mass was observed in the left maxillary region, 8x8x10 cm in size, firm and immobile, painless upon palpation, with no active bleeding.

This article was accepted: 28 May 2025
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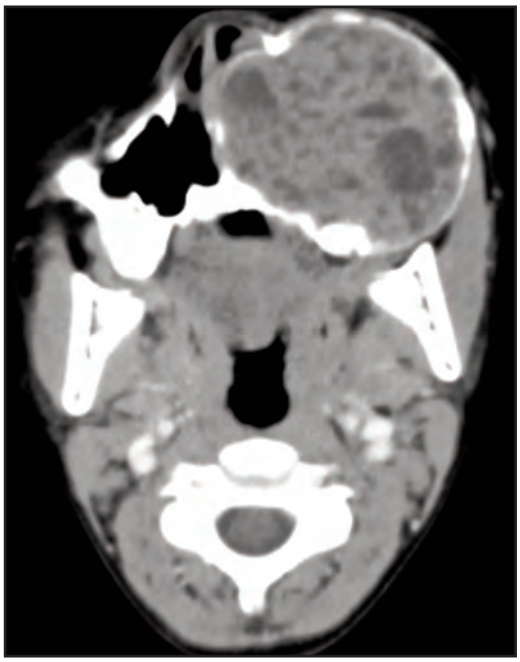

Fig. 1: Contrast CT scan of the head showing a mass arising from the left maxilla

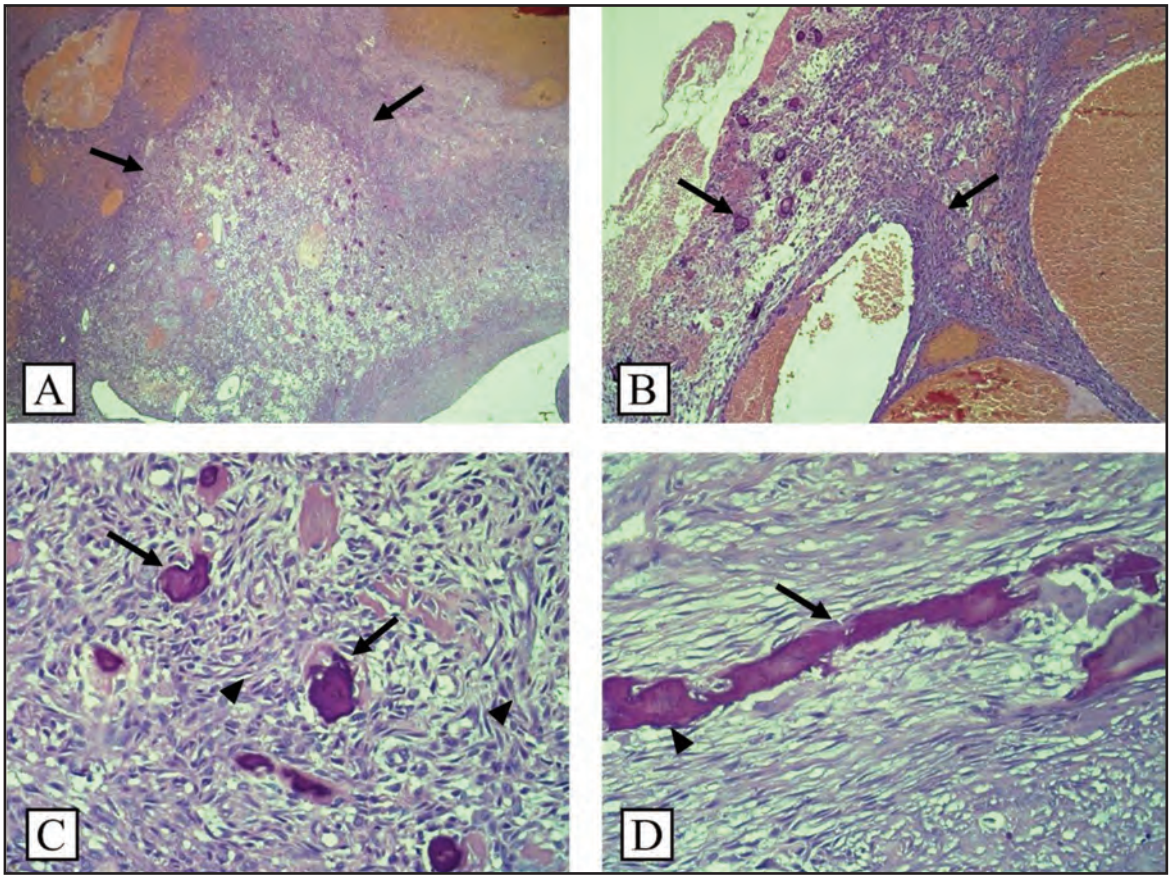

Fig. 2: Histopathology from surgical specimen stained with haematoxylin-eosin: (A), (B) fibro-osseous tumour mass (arrow) (4x, 10x); (C) ossification (arrow) between spindle cells (arrowhead) (10x); (D) bony trabeculae (arrow) with osteoblastic rimming (arrowhead) (40x)

Fig. 3: Preoperative (upper row) and postoperative image of the patient on 2-month follow-up (lower row)

Examination of the intraoral cavity showed a mass on the left maxilla pressing centrifugally and covering 30% of the oropharynx.

A CT scan showed a mass arising from the left maxilla suspected as an ameloblastoma, involving the 23, 24, 25, 26, 27, and 28 teeth, expanding to the left maxillary and ethmoidal sinus, nasal and orbital cavity, pressing onto medial and inferior rectus muscles causing left ocular proptosis, and destructing the left sphenoid wing, inferior orbital rim, maxillary and ethmoid sinus wall, and pterygoid plate. No intracranial involvement was observed. The lesion was accompanied by bilateral cervical lymphadenopathy, the largest being 1.2 cm in diameter. Biopsy of the lump showed ossifying fibroma.

After another month, the patient experienced a significant amount of bleeding from the lump and weight loss of eight kilograms. A treatment plan was devised together with a plastic and reconstructive surgery specialist to carry out a wide excision of the mass, followed by total maxillectomy and defect reconstruction using a free fibula flap. The total maxillectomy was performed using the anterior skin approach. The fibula free flap was anastomosed using end-to-end anastomosis between the donor peroneal artery and the recipient superior thyroid artery, and between the donor comitans vein and the recipient external jugular vein. The surgery was deemed a success, and the patient was instructed to return for postoperative follow-up.

On 2-month follow-up, the patient presented with good mastication and swallowing functions despite having no teeth on her left upper jaw and denied having any pain or difficulties with feeding, speaking, or walking. A skin paddle of the free fibula flap was observed to be the same colour as the donor with soft hairs. A left Weber-Ferguson incision scar and submandibular scar were in good condition.

DISCUSSION

Fibro-osseous lesions are rare benign tumours, with ossifying fibroma being the most common type in the head and neck region. JOF is a type of ossifying fibroma found in children, usually presenting as a aggressive, rapid-growing mass causing destruction to adjacent structures, and further classified into trabecular type (JTOF) and psammomatoid type (JPOF). JOF is most commonly found in the posterior mandible and very rarely arises from the maxilla.[1,2]

On a CT scan, JOF is characterised by a growing lesion with well-defined borders, is locally aggressive, and causes cortical destruction. The lesion might be radiolucent or radiopaque.[1] In our case, a CT scan showed an expansile lytic lesion with solid and cystic components arising from the left maxilla, causing cortical destruction without infiltration to the surrounding structures, a benign but aggressive lesion. On histopathology, JTOF is unencapsulated but has a well-defined border with a loose structure of hypercellularised stroma of spindle cells and osteoid structures in-between,

while JPOF is characterised by small uniform ossicles embedded in stroma of spindle and stellate cells.[3] In our patient, histopathology showed a fibro-osseous lesion, the fibrous component being proliferating fibroblasts with spindle cells, and ossifying component being cementum-like islands and osteoblastic rimming on bony trabeculae. No nuclear atypia or mitosis was found. Based on the results of anatomical pathology examination, specific classification between Juvenile Trabecular Ossifying Fibroma (JTOF) and Juvenile Psammomatous Ossifying Fibroma (JPOF) cannot always be determined in every case. Despite the presence of a large tumour, in some cases, the histological features distinguishing these two subtypes may not be sufficiently clear or adequately represented in the available tissue sample. However, based on the anatomical pathology findings, the histological features predominantly show ossicle or psammoma components rather than trabecular bone and its anastomosis. Therefore, the histological characteristics of this case are more consistent with Juvenile Psammomatous Ossifying Fibroma (JPOF).

Surgical management of OF depends on clinical and radiological manifestations. Titinichi5 proposed a management protocol in which curettage with peripheral ostectomy is the first-line choice of treatment, particularly for medium-to-large neoplasms of the maxilla and mandible, and those with well-defined borders on CT scan. Enucleation is reserved for small, well-defined lesions of the mandible. Resection with defect reconstruction is recommended for large, infiltrative lesions, particularly in the posterior maxilla; those with poorly defined borders and multilocular appearance on CT scan; or cases of recurrence. Resections should be done with a clear margin of not more than 5 mm.[3] In this patient, a wide excision was carried out considering the large size of the mass, its aggressive growth, and possibility of a recurrence, followed by a total maxillectomy and defect reconstruction using osteoseptocutaneous fibula free flap for maxillary framework and soft tissue palate.

Recurrence following conservative surgery (enucleation or curettage) is reported to be as high as 38.4%, compared to radical surgery (surgical resection) with a recurrence rate of 1.6%. Tumours with locally aggressive behaviour and ill-defined borders on CT have a higher recurrence rate compared to those with well-defined borders due to incomplete excision in the infiltrative borders.[4] In this patient, the wide maxillary defect was reconstructed using a free fibula flap, corresponding with studies by Carvalho et al.,[5] and Zhang et al.,[6] which reported complete excision as the main choice of treatment in OF with a high recovery rate and low recurrence rate. The studies also recommend defects caused by tumour removal to be reconstructed using free flap grafting.

A maxillary defect due to tumour removal or trauma might cause significant functional disruption and aesthetic deformity. The main objectives of reconstruction are to reduce deformity, restore function, particularly mastication and speaking, provide structural support for external facial reconstruction, and restore the external facial aesthetics. The free fibula flap was introduced by Hidalgo in 1989 and has been a widely used method of maxillofacial reconstruction.[6] Reconstruction in paediatric patients has been shown to provide satisfactory results with minimal postoperative morbidity.

The osteoseptocutaneous fibula free flap provides bone, muscle, and skin that can be utilised simultaneously in a defect reconstruction. It has good periosteal blood supply, facilitating multiple osteotomies and allowing the fibula to be moulded according to the desired maxillary contour, and allows the possibility of dental implants in the future. The free fibula flap is also superior for reconstruction with a large defect, providing adequate tissue while maintaining the "like with like" principle. The fibula itself is a non-weight-bearing bone and can be harvested up to 25 cm in length, leaving only the most proximal and distal parts to preserve the knee and ankle joints.[7] However, the fibula flap does come with certain limitations. Fibula free flap in oncology patients may have cause delays in starting adjuvant treatments, such as radiotherapy, due to the postoperative recovery period.[8] Paediatric patients undergoing the surgery will also require long-term monitoring to assess their growth, such as craniofacial growth and gait.[9]

Postoperative complications at 30 and 90 days following free flap surgery in paediatrics were reported in less than 20% of patients, consistent with other studies reporting minimal complications following free flap reconstruction in maxillofacial defects.[2] Slijepcevic et al.,[9] reported free flap failure incidence in only 1 out of 89 patients of paediatric maxillomandibular reconstruction with fibula free flaps, showing that reconstruction using free flaps can be utilised in paediatric patients with a high success rate and low risk of failure. The most common postoperative complications were wound healing issues, such as cellulitis and wound dehiscence.[2] On 2-month follow-up, our patient had good mastication and swallowing functions despite having no teeth on her left upper jaw and denied having any pain or difficulties with feeding, speaking, or walking. Long-term outcomes and complications are evaluated using questionnaires of patient satisfaction and quality of life. In our centre, we use a questionnaire that has been translated and validated into the Indonesian language.

Long-term complications related to craniofacial growth were reported in a quarter of patients and mostly occurred in patients younger than 10 years of age or with lateral or hemimandibular defects.[9] The satisfying result is also seen in the long-term outcome of osteoseptocutaneous fibula free flap for various extended mandible defects, both in the function and quality of life (QoL). The QoL showed satisfactory outcome in swallowing, speech, eating function, and also donor site morbidity. The neo-mandible also seems to be well-developed and symmetrical despite the lack of a growth point.[10] Dental rehabilitation may facilitate normal occlusal relationship and midface growth.[9] As of the writing of this article, our patient has been referred to a prosthodontist for a dental rehabilitation.

Management of patients with JOF is best carried out by a multidisciplinary group comprising head and neck surgeons, plastic surgeons, radiologists, anaesthesiologists, and nursing staff experienced in head and neck cancer reconstruction. Other professionals might be added into the team if deemed necessary.

CONCLUSION

JOF is a rare fibro-osseous lesion characterised by an aggressive nature and a high recurrence rate. Diagnosis should be based on a combination of clinical-pathological and radiological correlations. Management of JOF requires a balance between extensive lesion removal while preserving the surrounding anatomical structures. In paediatric oncological cases requiring maxilla-mandibular reconstruction, the free fibula flap offers significant benefits by providing a bony framework and replacing the soft tissue components such as the palate, gingiva, and buccal mucosa. This approach helps minimise long-term complications, including plate extrusion, infection, and growth disturbances in children. Multidisciplinary teamwork is important in managing these patients to ensure the best outcomes.

ACKNOWLEDGEMENT

None.

DECLARATION

Permission was obtained from the parents for this report. No funding was received for this manuscript. There was no conflict of interest to be declared.

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