Volume 3 / Issue 1 / June 2023

Clinical Report

Surgical and Prosthetic Management of a Juvenile Ossifying Fibroma In a 5-Year-Old Child: A Clinical Report

Bulent Piskin, DDS, PhD[1], Abdullah Durmaz, MD[2], Cumhur Sipahi, DDS, PhD[3], Alper Uyar, DDS, PhD[3], Savaş Özarslantürk, DDS, PhD[4*], Nesrin Şahin, DDS, PhD[1], Ferhat Cüce, MD[5].

[1]Cappadocia University, Faculty of Dentistry, Prosthetic Dentistry, Nevşehir, Türkiye
[2]University of Health Sciences, Faculty of Medicine, Otolaryngology, Head and Neck Surgery, Ankara, Türkiye
[3]University of Health Sciences, Faculty of Gulhane Dentistry, Prosthetic Dentistry, Ankara, Türkiye
[4]University of Health Sciences, Faculty of Gulhane Dentistry, Dentomaxillofacial Radiology, Ankara, Türkiye
[5]University of Health Sciences, Faculty of Gulhane Dentistry, Radiology, Ankara, Türkiye
**Corresponding author: savasozarslanturk@yahoo.com*

Piskin, Bulent., Durmaz, Abdullah., Sipahi, Cumhur., Uyar, Alper., Özarslantürk, Savaş., Şahin, Nesrin. & Cüce, Ferhat. "Surgical and Prosthetic Management of a Juvenile Ossifying Fibroma In a 5-Year-Old Child: A Clinical Report". *Acta Stomatologica Cappadocia.* 3;1 (June 2023): 51-61
DOI: https://doi.org/10.54995/ASC.3.1.5

Received: 25.04.2023; Accepted: 09.05.2023

Surgical and Prosthetic Management of a Juvenile Ossifying Fibroma In a 5-Year-Old Child: A Clinical Report

Bulent Piskin, Abdullah Durmaz, Cumhur Sipahi, Alper Uyar, Savaş Özarslantürk, Nesrin Şahin, Ferhat Cüve

Abstract

Juvenile ossifying fibroma (JOF) is a rare and benign fibro-osseous lesion of the craniofacial skeleton commonly seen in children under 15 years of age. Despite being classified as benign, JOF lesions are aggressive in nature and reach large dimensions in a short period of time. Thus, their resections may cause large orofacial defects which have serious detrimental effects on the functional and nutritional requirements of pediatric patients being in the period of growth. Therefore, its early diagnosis and proper surgical and prosthetic management are of vital importance for the survival of patients. However, no detailed reports in the literature describe the prosthetic management of large intraoral defects in pediatric patients under 10 years old.

Keywords: Juvenile Ossifying Fibroma, Maxillary Defect, Prosthetic Management, Obturator, Pediatric Obturator, Surgical Pediatric Obturator, Interim Pediatric Obturator

Introduction

Juvenile ossifying fibroma (JOF) is a rare and benign fibro-osseous neoplasm of craniofacial skeleton characterized by early starting age and highly specific location.[1-3] These lesions are commonly seen in children under 15 years of age and paranasal sinuses, mandible, maxilla, orbital and fronto-ethmoid bones are the most commonly affected structures.[1,4-6] Despite being classified as benign, JOF lesions can show a highly aggressive clinical course due to their rapid growth potential, excessive osteoblastic activity, and high recurrence rates.[1,2,5,7] Thus, they may imitate malignant tumors and reach large dimensions in a short period.[1,2] Additionally, painless progress of the lesions may delay the diagnosis[8], and voluminous jaw defects may occur as a result of repetitive surgical interventions in case of recurrence. Inevitably, these defects disturb the nutritional requirements of pediatric patients in the period of growth and development.[9]

Surgical, orthodontic, and prosthodontic management of cleft palate and other congenital defects have been well described previously. In addition, the management of maxillofacial

neoplasms in child patients older than 6 years old is the subject of numerous studies in the literature. However, the vast majority of existing studies emphasized the surgical aspects of rehabilitation, and the presented data are not sufficient in terms of prosthetic management.

The aim of the present report was to describe the detailed management of a recurrent maxillary JOF lesion in a 5-year-old child.

Case

A 5-year-old girl was admitted to the Otolaryngology, Head and Neck Surgery Clinic with a complaint of nasal obstruction and infraorbital swelling. Endonasal endoscopic examination of the patient revealed total obstruction of the right nasal passage and gradual narrowing of the left passage. A large fibro-osseous lesion invading the right maxillary sinus was observed by computed tomography evaluation. The lesion was resected with wide margins by nasal endoscopic approach and diagnosed as JOF after its histopathologic evaluation.

The lesion recurred in a very short period of time, and medial maxillectomy was performed with the endonasal endoscopic approach. However, the patient was presented with similar complaints 10 months after the medial maxillectomy operation. Clinical and radiographic examinations revealed a large recurrent lesion invading the entire right maxillary sinus. The recurrent lesion reposed on the pterygoid plate of the sphenoid bone posteriorly, the crestal bone of the maxilla anteriorly, and the inferior border of the right orbita superiorly (Figure 1).

Figure 1. Computed tomography image of the recurrent lesion.

Therefore, it was decided to perform the resection of the right maxilla via transoral approach and a surgical obturator (SO) had to be made before the resection of the maxilla in order to separate oral and nasal cavities throughout the healing period.

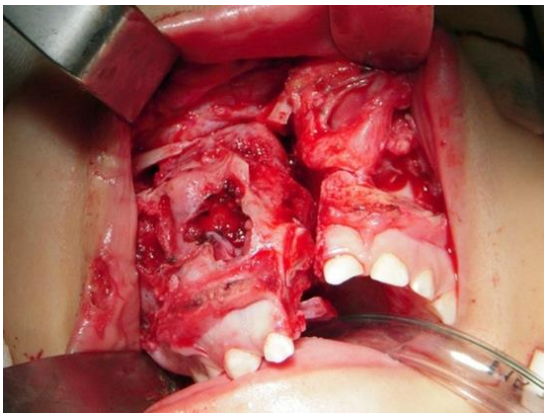
Planning and fabrication of the SO

Impression of the maxilla was made with irreversible hydrocolloid impression material (PhasPlus, Zhermack, Badia Polestine, Italy), and the master model was obtained with type III dental stone (Moldano, Heraeus Kulzer, Hanau, Germany). The estimated cavity of the post-resectional defect was formed on the master model with the guidance of CT images. An Adams clasp was placed on the maxillary second primary molar and a vestibular arch was prepared from round stainless-steel wire (Remanium, Dentaurum, Ispringen, Germany) for the retention of the obturator. The surface of the cavity was isolated (Isolant/C.M.S, Dentsply, UK), and a layer of auto-polymerizing acrylic resin material (AARM) (Ortocryl, Dentaurum, Ispringen, Germany) with a thickness of 2 mm was applied in order to set an acrylic hollow bulb. The obtained acrylic cavity was filled with pink modeling wax (Cavex Set Up, Regular, Haarlem, Netherlands), and a second layer of AARM in the same thickness was applied onto wax surfaces and the left side of the master cast. Thus, a hollow bulb was formed and integrated with the base of the SO with clasps incorporated. Eventually, the remaining modeling wax was eliminated from the bulb via a small aperture with the aid of hot water and finishing procedures were performed before surgical intervention.

Resection of the affected maxilla and insertion of the SO

The whole tumoral mass, right hard palate, alveolar bone with relevant dentition except the right central incisor, right maxillary sinus, and right maxillary tuberosity were resected with radical transoral hemimaxillectomy (Figure 2).

Figure 2. Frontal view of resected maxilla.



The SO was inserted into the mouth and fitted on the remaining tissues with minimal pressure in order to protect the incision and suture lines. Tissue conditioner material (TCM) (Visco-gel, Dentsply, Konstanz, Germany) was used to seal the small gaps between the obturator and remaining tissues and periodically renewed with 3 days intervals (Figure 3).

Figure 3. Intraoral view of the surgical obturator (SO).

No complications have been observed at the end of three weeks healing period. However, the parents revealed that SO did not meet the functional needs of the child. This situation was also confirmed by intraoral examination. We observed that the child's upper lip movements had caused unfavorable forces which were directly transferred onto the vestibular arch of the SO. It was determined that this retention element provoked loss of retention paradoxically. Therefore, a delayed surgical obturator (DSO) which did not include any vestibular arch was fabricated and delivered.

Fabrication of Interim Pediatric Obturators (IPO)

At the end of the quarterly healing period, the remaining intraoral and nasal tissues were healthy enough and these conditions encouraged the treatment team to fabricate an IPO. Due to extreme anxiety arising from ongoing interventions, general anesthesia was administered to the child for making an impression. Undercuts in the defect area were blocked out with vaseline-laden gauze, maxillary and mandibular impressions were made and master casts were obtained. An acrylic hollow bulb was fabricated on the maxillary master cast as described previously. Afterward, a metal framework that could be able to fulfill the functional requirements of the IPO such as sufficient retention, stabilization, and comfort was planned. The major connector of the framework was a semi-palatal U plate seating on the healthy remaining hard palate. Three Akers clasps were placed on the remaining primary canine and molars. The planned framework was cast from a base metal alloy (Remanium CSe, Remanium, Ispringen, Germany) and its fit was verified intraorally. Finally, the base of the metal framework was extended using AARM and prepared for the tooth setting.

Due to the limited marketing of pediatric artificial denture teeth (PADT), a set of PADT (Bambino, Major Prodotti Dentari, Torino, Italy) with the closest appearance to existing teeth was selected. The registration of the intermaxillary relationship was not possible due to the limited mouth opening. Thus, the tooth setting procedure was performed using the guidance of the opposing mandibular arch and interocclusal harmony was directly adjusted intraorally. Try-in procedure was completed, the master model was flasked and the IPO was processed using heat-polymerized acrylic resin (Meliodent, Heraeus Kulzer, Newbury, Berkshire, UK). Following the finishing and polishing procedures, the IPO was delivered to the patient with the recommendations for usage and daily care (Figure 4).

Figure 4. Intraoral view of the Interim Pediatric Obturator (IPO).

During periodical controls, the patient and her parents complained about insufficient retention of the IPO. Additionally, chronic inflammation was observed on the palatal mucosa which was covered by the base of the IPO. It was also seen that initially sound maxillary right primary central tooth become mobile and its periodontal support was dramatically decreased. Thus, it was extracted and a new and more retentive IPO was planned.

Once again, impressions were made and master casts were obtained as described previously. The major connector of the first IPO was redesigned to protect palatal tissues. To increase retention of the IPO, a modified multiple circumferential clasp assembly was placed on all remaining primary teeth as a direct retainer. However, the labial bar, 17 mm in length, 5 mm in width, and 0.95 mm in thickness, was the most striking component of the new framework. It extended from the mesial surface of the maxillary primary central incisor to the distal surface of the left primary lateral tooth parallel to the attached gingiva of the anterior teeth. The 2nd IPO was fabricated and delivered to the patient with the previously described technique (Figure 5).

Figure 5. Intraoral view of the 2nd Interim Pediatric Obturator (IPO).

Intraoral examination findings and CT images of the remaining maxilla did not reveal any sign of recurrence at the end of six months. In addition, soft tissues surrounding the defect cavity were completely healthy and properly epithelialized. Despite increased retention and stabilization, the appearance of the 2nd IPO was not found satisfactory by the patient who has attended elementary school. Additionally, the maxillary permanent incisor tooth was mobile due to the eruption of the permanent. Therefore, the 3rd IPO was fabricated with the same design but adult ADT was preferred instead of pediatric ones in order to achieve proper appearance and shade matching, after the eruption of the maxillary primary incisor tooth (Figure 6).

Figure 6. Intraoral view of the 3rd Interim Pediatric Obturator (IPO).

After a follow-up period of 3 months, in which no serious complaint was declared, gradually increased retention was reported by the patient's parents. Intraoral soft and hard tissues were found completely healthy. Thus, it was decided to keep the 3rd IPO until the eruption of permanent first molars.

Discussion

Rapidly growing lesions of the craniofacial skeleton such as JOF could have a strong potential for recurrence even if they did not include malignancy.[1,4] Thus, early diagnosis and management are of vital importance to avoid the occurrence of large orofacial defects. Because the main symptoms of JOF lesions such as facial swelling and asymmetry are closely related to the field of interest, dentists may play a crucial role in early diagnosis.

Adequate nutrition should be provided to pediatric cancer patients to get the maximal outcome from the treatment and to support healthy growth and development.[9] However, intraoral defects cause serious problems in chewing and swallowing functions, thus their prosthetic management becomes highly crucial, especially in the growth and development period. Despite its importance, only a few papers have focused on the prosthetic rehabilitation of pediatric patients with maxillofacial defects.[10,11] However, no report was found in the literature about prosthetic management of maxillary defects in patients under 6 years of age. Existing few reports differed from the present case with regard to the age of patients.

The usage of an interim obturator in a 10-year-old child, who underwent partial maxillectomy due to squamous cell carcinoma, was reported in one of these studies. However, it did not provide any detailed information about its fabrication technique.[10] Dominguete et al[11] reported prosthetic management of a mandibular defect using dental implants, which occurred by the radical resection of a JOF lesion. However, the patient rehabilitated by the authors was much older (18 years old) than the present patient, and making a comparison between the two treatment methods seems almost impossible.[11]

In fact, practitioners who attempt to rehabilitate the intraoral defects in pediatric patients in primary dentition may experience some difficulties when compared with older ones with permanent dentition. Making an impression was one of the most challenging stages of the present case due to the severe anxiety of the child arising from subsequent surgical and clinical interventions. Thus, all impressions were made while the patient was under general anesthesia. Establishing precise intermaxillary relations was also difficult not only because of severe anxiety but also the destruction of tissue integrity. However, the most challenging issue was to ensure sufficient retention and stabilization for obturators in the present case due to the indequate depth of undercuts of primary teeth. To obtain sufficient retention and stabilization for the SO, not only the frictional property of the TCM but also retentive aspects of the Adams clasps and vestibular ach were planned to be effective. However, the vestibular arc was far from providing additional retention due to the morphology of the primary teeth. It was observed that the vestibular arch transferred compressive loads arising from upper lip movement and caused introral discomfort due to its harmful compressive load to the upper lip. Therefore, it was thought

that the use of vestibular arc in pediatric SO rehabilitating the maxillary defects might not be suitable for retention. Indeed, sufficient retention and stabilization could be obtained with DSO supported by the frictional effect of TCM.

It was expressed by the parents of the patient that the 1st IPO with cast clasps was more comfortable than SO and DSO. However, some retention and stabilization problems were confronted during the usage of the 1st IPO. The TCM, which the patient got used to the convenience provided by it, could not be utilized with the 1st IPO owing to the application difficulties and risk of candida infections. Thus, it could not be benefited from the frictional effect of the TCM which provides increased retention for the obturators. Under these circumstances, a special prosthetic design not formerly used in a pediatric defect patient was planned for the 2nd IPO. In fact, it was observed that continuous multiple circumferential clasp assembly was highly useful to provide maximal retention from the minimal curvature of the primary teeth. However, the most distinct aspect of the 2nd IPO was the bar located on the buccal mucosa of the remaining anterior teeth. The benefit of this substructure was observed during clinical trials such as try-in steps. It was expressed by the patient that the new prosthetic design provided sufficient and satisfactory oral comfort. However, the expressed outcomes of the 2nd IPO may not only be due to its structural difference but they may also be attributed to its application period. Surely, the contact between the acrylic parts of the IPO and the residual tissues, such as the hard palate and defect walls, was more concordant than the 1st IPO, due to the completed wound healing. Thus, its fabrication time might have been more suitable for using the 2nd IPO.

The greatest difficulty of the present study was to solve the functional and aesthetic problems with just an obturator. The aesthetic demands of the patient grew substantially after she attended primary school. More effort had to be spent to reach a natural appearance to prevent social and emotional maladjustments.

The growth and development of the craniofacial structures are firmly interrelated with each other[12] thus, existing defects affect the facial growth of the patient inevitably. Maximal changes in face growth (approximately 80% of total change) occur in children between 5 and 15 years old and the growth spurt starts early in females than males.[13] Therefore, substantial effects of maxillary resection on midfacial growth can be expected in the present case. However, all obturators used in the present study may prove right an old paradigm called “work with growth”[12] and can minimize probable effects on facial growth as much as possible. Thus, the final plan of the treatment team was to minimize the probable effects during the growth and development stage and contain the defect dimensions using growth spurts until the surgical management of the defect site.

Clinical and radiographic examination of the defect cavity and surrounding tissues did not reveal any recurrent lesion at the end of a follow-up period of 6 months. No pathological signs around the periodontal tissues of the remaining teeth were detected. Nevertheless, not any serious complaint was reported by the patient and her parents except for excessive salivation.

Undoubtedly, prosthetic management of defective maxilla in a child patient involves some difficulties such as inadequate retention and stabilization of obturators arising from insufficient contoured morphology of primary teeth. Therefore, the treatment team should be knowledgeable, open-minded, and creative to implement the new prosthetic design features in these cases.

References

1. Rinaggio J, Land M, Cleveland DB. Juvenile ossifying fibroma of the mandible. J Pediatr Surg 2003;38:648-50.
2. Breheret R, Jeufroy C, Cassagnau E, Malard O. Juvenile ossifying fibroma of the maxilla. Eur Ann Otorhinolaryngol Head Neck Dis 2011;128:317-20.
3. Zama M, Gallo S, Santechhia L, Bertozzi E, et al. Juvenile active ossifying fibroma with massive involment of the mandible. Plast Reconstr Surg 2004;113:970-4.
4. Tortirici S, Buzzanca ML, Burruano F, Difalco B. Juvenile central ossifying fibroma of the mandible: a case report. J Pediatr Surg 2010;45:e1-4.
5. Cicciu M, Herford AS, Juodzbalys G, Cicciu D. Juvenile ossifying fibroma of the maxilla: A rare aggressive case in a young patient. J Cancer Res Ther 2013;9:324-8.
6. Linhares P, Pires E, Carvalho B, Vaz R. Juvenile psammomatoid ossifying fibroma of the orbit and paranasal sinuses. A case report. Acta Neurochir (Wien) 2011;153:1983-8.
7. Zawadzka-Glos L, Brozek-Madry E, Chmielik M, Brzewski M, et al. Aggressive psammomatoid ossifying fibroma in a 3-month-old boy-A case report. Int J Pedaitr Otolaryngol Extra 2011;6:143-5.
8. Zawadzka-Glos L, Brozek-Madry E, Chmielik M, Brzewski M, et al. Aggressive psammomatoid ossifying fibroma in a 3-month-old boy-A case report. Int J Pedaitr Otolaryngol Extra 2011;6:143-5.
9. Bauer J, Jürgens H, Frühwald MC. Important aspects of nutrition in children with cancer. Adv Nutr 2011;2:67-77.
10. Binahmed A, Charles M, Campisi P, Forte V, et al. Primary squamous cell carcinoma of the maxillary alveolus in a 10-year-old girl. J Can Dent Assoc 2007;73;715-18.

11. Dominguete PR, Meyer TN, Alves FA, Bittencourt WS. Juvenile ossifying fibroma of the jaw. Br J Oral Maxillofac Surg 2008;46:480-1.
12. Enlow DH, Hans MG. Essentials of facial growth. 1st Ed. WB Saunders Company. Philadelphia; 1-98:1996.
13. Bishara SE. Facial and dental changes in adolescents and their clinical implications. Angle Orthod 2000;70:471-83.