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Surgical Management of Recurrent Ossifying Fibroma: A Case Report

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Abstract: ***Introduction: Ossifying fibroma (OF) is one of the so-called fibro osseous lesions, consisting of a benign fibro-osseous lesion characterized by slow growth and proliferation of fibrous cellular tissue, bone, cement or a combination, occurs exclusively in the tooth bearing areas of the jaws. The aim is to report arecurrent case of ossifying fibroma in pediatric patients under surgical management. Case report: A6 years old girl patient with chief complain of a lump on her right lower jaw, with painless, firm, and non bleeding characteristic on the clinical findings. Panoramic x-ray showed irregularly-shaped radiopaque intrabony lesion with ill-defined margins. The patient went for excisional biopsy and extraction of deciduous teeth,with pathological result was ossifying fibroma. However the lesion recurred after 8 months later, and the second surgery was done with re-excision of the tumor. There has been no sign of recurrence since the last surgery and we have continued follow-up observation. Conclusion: Ossifying fibroma is a benign fibro-osseous lesion with various rate of recurrency, surgical is a recommended management for the lesion, followed by periodically observation.***

Keywords: Ossifying fibroma, recurrent, pediatric, surgical

1. Introduction

Ossifying fibromas are benign asymptomatic neoplasms of the jaw that generally have slow growth and present proliferation of fibrous cell tissue, with a varying quantity of bone products that include bone, cement or a combination of these.[1] They are often considered in to the broader category of benign fibro-osseous lesions (BFOLs). All BFOLs are characterized by the replacement of native bone by fibrous and mineralized tissues and are grouped together due to their histologic similarities despite having different clinical features and treatments. The other BFOLs, fibrous dysplasia and the cemento-osseous dysplasia, can be difficult to differentiate microscopically, and therefore all BFOLs require radiographic and clinical correlation to make an accurate diagnosis.[2]

Ossifying fibromas occur most often in the posterior region of the mandible and may also occur in the maxilla, commonly in the region of the canine fossa and in the area of the zygomatic arch. They are more common in females, and present greatest incidence in the third and fourth decades of life. Facial asymmetry and tooth displacement may occasionally occur.[1] Upon radiographic examination, it is observed that the edges of the lesion are usually well defined, with a thin radiolucent line that represents a fibrous capsule. The internal structure shows mixed radiolucent–radiopaque density, with a pattern that depends on the form and quantity of the calcified material that is present. The differential diagnosis is generally made with other lesions that present mixed radiolucent–radiopaque internal structures, especially with fibrous dysplasia.[3]

Because it is believed to be a neoplasm, the treatment is surgical; in fact, the lesions often shell out easily at surgery, although there is a recurrence rate that has variously been reported from 1% to 63%. For these reasons, some authors recommend aggressive treatment for more aggressive lesions, including aggressive curettage, localized surgical resection, and segmental mandibular resection. When present in the craniofacial complex, treatment may have to be more aggressive to protect the vital structures.[4] The aim ofthis study was to report on a clinical case of recurrent ossifying fibroma on pediatric patient after 8 months that was treated by means of the tumor excision.

2. Case Report

A 6 years old girl patient came with chief complain of a lump on her right lower jaw. About 8 months prior to admission, the patient complained of small lump peanut size on her right lower gum with no pain or bleeding and same color with surroundings gum so she didn't seek any treatment. Five months later she complained the lump was getting bigger, then she was referred to Hasan Sadikin Hospital Oral and Maxillofacial Surgery Dept. for further treatment. On physical examination, the patient had normal vital signs. Clinically, a marble size lump was noted over her right mandible which was firm and non-tender on palpation (Fig.1a). From the intra oral, the tumor was found on the buccal side of the right mandible obliterating the gingiva, alveolar bone, and mucobuccal fold over the region of tooth 82-84 (Fig.1b). The mass was firm and not tender on palpation. Panoramic x-ray showed irregularly shaped intrabony lesion with ill-defined margins extending from the region of teeth 82-84 (Fig.1c); radiopacity were seen with the radiolucent area indicating increased calcification of the tumor.

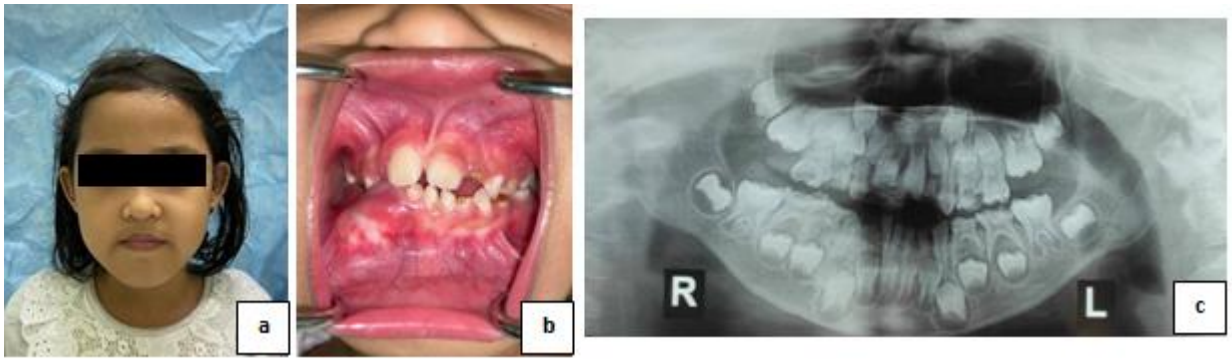

Figure 1: (a) Extraoral and **(b)**intraoral findings and **(c)** panoramic x-ray was obtained

The lesion was managed with excisional biopsy (Fig.2a) followed by extraction of teeth 82,83,84 (Fig.2b), with a pathological result showed the microscopic features of ossifying fibroma (Fig.3).

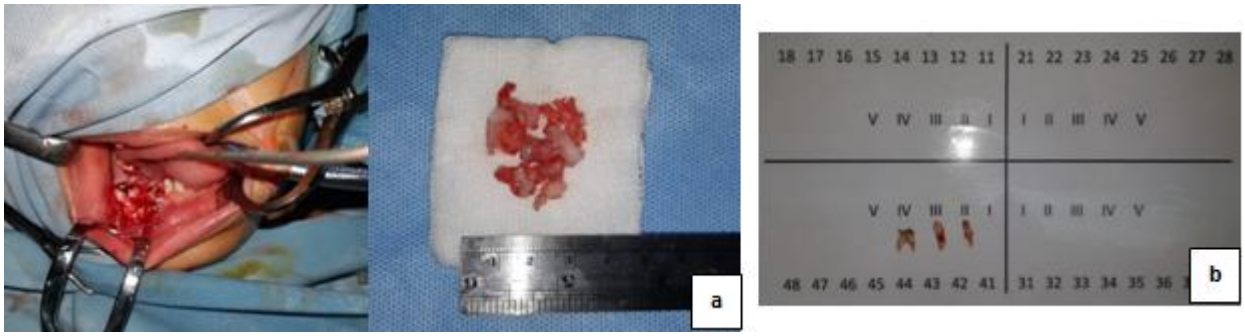

Figure 2: (a) Excisional biopsy followed by **(b)** extraction of teeth 82,83,84

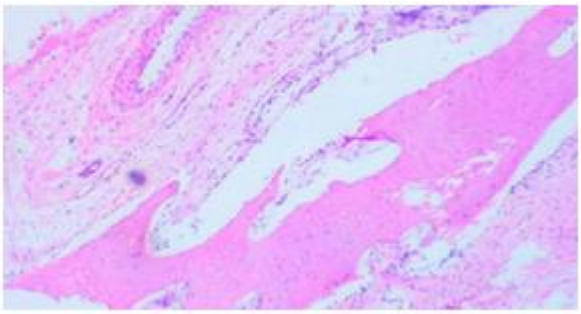

Figure 3: Microscopic view of bone trabecular ossified with osteoblastic rimming

e patient was observed periodically to examine if there was any recurrence of the lesion. However, 8 months laterit was found the lesion recurred at the same site with the same clinical findings before the first surgery (Fig.4a,b). Panoramic x-ray was obtained with irregularly shaped intrabony lesion diffuse margins on the previous operation site where radiopacity were mixed within the radiolucent area (Fig.4c)

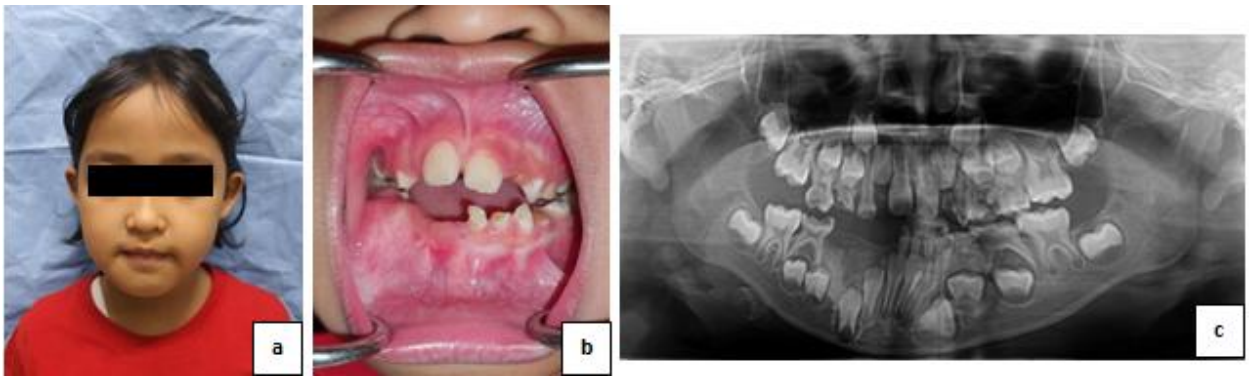

Figure 4: (a,b) Recurrence lesion on both clinical and **(c)** radiograph findings

Considering growth age of the patient and the parents was well informed, it was decided to perform re-excisionof the lesion (Fig.5a,b) and extraction of tooth 85 (Fig.5c).

Figure 5: (a,b) Re-excision of the lesion and **(c)** post operative view

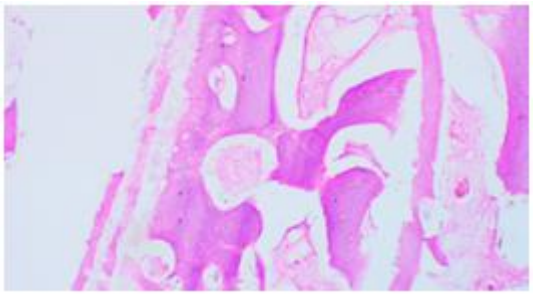

Figure 6: Pathological result of mature bone trabecular with osteoblastic rimming

The histopathologic examination shows formation of bone trabeculation within fibrous connective tissue with osteoblastic rimming being strongly evident at periphery of the trabeculae (Fig.6). These findings confirmed the diagnosis of ossifying fibroma. There has been no sign of recurrence since the last surgery and we have continued follow-up observation.

3. Discussion

Maxillofacial fibro-osseous lesions comprise a group of face and jaw disorders characterized by the replacement of normal bone by a benign connective-tissue matrix with varying amounts of mineralized substances. The term of "fibro-osseous lesion" is not a specific diagnosis and describe only a process. Ossifying fibroma as known as cemento ossifying fibroma is classified into fibro-osseous and osteochondromatous lesion, and is also categorized into benign mesenchymal odontogenic tumors. OF There were three clinicopathological variants of OF, which have been identified as OF, juvenile trabecular ossifying fibroma (JTOF) and juvenile psammomatoid ossifying fibroma (JPOF).[5]Epidemiology, it has proposed that the peak incidence of OF has been in the third and fourth decades of life. Furthermore, there is a definite female prediction with a ratio as high as 5:1. OF occurs exclusively and more commonly in the tooth-bearing area of the mandible rather than that of maxilla, and the most common site is mandibular premolar and molar area.[6]

Some authors mentioned that the OF occurring at maxilla behaves more aggressively with more obvious symptoms and signs than those in mandible. It may result from the different physiological natures between mandible and maxilla. The mandible contains a thicker outer cortex with loose inner marrow, and the thick cortex is a barrier for the growth and expansion of the lesion. On the contrary, the lesion in maxilla is easier to expand in thin cortex and inner cancellous bone.[7]In this report, in a 6 years old girl patient with the lesion occurred in the right mandibular parasymphysis and mainly involved mainly the buccal cortex area, interestingly this OF lesion occurred in pediatric which according to our knowledge JTOF was the most likely to occurred during this age.

The clinical features vary depends on cases. Generally, OF presents as a painless expansion,large tumor expands maxillary sinus or inferior border of the mandible, displacement of teeth may be the only early clinical feature, as it showed on deciduous teeth 82,83,84 of this patient. The lesion is therefore frequently ignored by the patient until the growth produces a noticeable swelling and facial deformity.Radiographically, it appears typically as a radiolucent lesion in an early stage. However, over timeas the tumor matures, there is increasing calcification so that the radiolucent area becomes flecked with opacities until ultimately the lesion appears as an extremely radiopaque mass. The OF presents a radiolucent appearance in 53%, a sclerotic radio density in 7% and mixed or mottled appearance in 40% of the cases.[8]In our case, the patient had painless and bony hard swelling on the right mandibular parasymphysis without numbness. The images showed that the lesion was not wellcircumscribed and had mixed radiopaque fleck with surrounding radiolucent area.

Histopathologically, the more radiolucent lesions are composed of cellular fibrous connective tissue, frequently in a whorled pattern. Collagen fibers are often arranged irregularly, although a whorled, uniform pattern may be evident. Calcified deposits are noted throughout the fibrous stroma. The nature of the hard tissue is generally quite variable within a given tumor as well as between lesions. Irregular trabeculae of woven bone or lamellar bone are most consistently noted in these tumors.[7] In this case both of the first and second pathological result showed OF designation with ossified bone trabecular including osteoblastic rimming. Whereas those reported in the case of JTOF has showed similar findings but with high vascularity that makes it more aggressive.[8]So this case had no finding to reveal juvenile aggressive OF. While the etiology has yet been unknown, trauma may act as a predisposing factor, which suggest a connective tissue-reactive etiology rather than a neoplastic one.[7]In this patient there was no history of trauma and injury of facial region.

Some authors have reported that expansive recurrent lesion after the surgery and untreated massive tumors may require resection, with the recurrence rate of OF were from 12 % to 28 %.[5]Treatment of OF generally has been varying from conservative to radical surgery, depending on the size and location of the individual lesion. Radical treatment of the tumour such as an en bloc resection should be considered if there are recurrences due to its aggressive nature.[8] According to a review by Liu et al, OF requires radical surgery because the possibility of malignant transformation. All reported patients in their study, with partial or incomplete resection experienced recurrence. The time of recurrence was always unpredictable, ranging from 6 months to 7 years after the operation in our reports. Therefore, there must be a long enough follow-up period of at least 10 years. In another recurrent cases in the study, the tumor often became much larger or the radiographic appearance changed extensively. However it is known, most OF once completely excised, do not recur.[9]Based on these findings and knowledge, we planned the treatment of this patient with an intraoral approach excision of the lesion.The management of tooth involved in this kind of lesion has also been controversial.[7] As a resultof the second surgery we finally needed to remove four teeth in this case. The recurrence in this case might be developed due to residual lesion from an incomplete first excision. Therefore re-excision was perfomed on the second surgery with more aggressive in order to avoid any future recurrence.

4. Conclusion

As the conclusion of this paper, the clinical, radiographic and pathological findings of a OF should be considered altogether in order to determine the correct diagnosis and treatment. Due to its high recurrence rate, radical treatment of the OF should be considered to avoid any future recurrence, any further complication of patient's cosmetic and functional problems. In addition, all treated OF cases should be strictly followed up.

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