Case Report

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Trabecular Type Juvenile Ossifying Fibroma Of Mandible

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1. Abstract

Juvenile ossifying fibroma is a benign lesion with aggressive local growth. It is similar to an ossifying fibroma, although it is more aggressive and of earlier onset. The tumor mainly occurs in juveniles, majority of the patients being 15 years or younger. Radiologically as well as at surgery, the lesion is well demarcated from its surroundings. In this article, we report a case of juvenile ossifying fibroma of mandibular anterior region extending from canine to canine region with expansion of both buccal and lingual cortical plates with a history of 6-month duration in an 8-year-old girl. The treatment was complete enucleation of the tumor mass and curettage of the bony cavity with primary closure. The patient is on regular follow-up of2 years with no recurrence.

2. Introduction

Ossifying fibroma, a rare tumor entity, is a well- demarcated benign fibro-osseous tumor with capsule composed of metaplastic bone, fibrous tissue and varying amounts of osteoid [1,2]. Ossifying fibromas are subdivided into conventional and juvenile clinico-pathologic subtypes [1]. Conventional ossifying fibromasare usually slow growing and generally seen in the third and fourth decades of life [3]. They are treated withsimple curettage, and the recurrence is rare [4]. It affects people of all ages, but in contrast to the form seen at adults, the juvenile form is clinically more aggressive and tends to be recurrent [1].

Juvenile ossifying fibroma (JOF) has been distinguished from the rest of ossifying fibromas by the age in which it appears (younger than 15 years old), the zonethat is involved and its clinical behavior [5]. The tumor is normally asymptomatic, reaching a great size and being locally aggressive. JOF appears at an early age; 79% of the patients arediagnosed before the age of 15 [1,6,7]. Males and females are equally affected8. JOF originates from periodontal ligament and ranges 2% of oral tumors in children9. The JOF is located mainly (85%) in facial bones, in some cases (12%) in calvarium and very seldom (3%) extracranially [6]. Ninety per cent of the lesions located inthe face region involve the sinuses, mainly the maxillary antrum. Mandibular lesions are seen in 10% of the cases [6,10]. The tumor is well circumscribed by a tiny sclerotic shell of bone. It appears locally aggressive withcortical disruption and involvement of many adjacent anatomical structures. This lesion has predominating soft-tissue consistency with variable amounts of internal calcification and/or linear or irregular focal bone [6].

It usually shows a low-density mass due to cystic changes on computed tomography (CT) scans. Following intravenous injection of iodinated contrast, the lesion may show diffuse appearance enhancement {6}. Histologically, JOF is characterised by the presence of cellular fibrous stroma, garland-like bony strands and cement particles [3,6,8,9]. JOFs are classified into two distinct clinicopathological entities: the trabecular and the psammomatoid types. Trabecular JOF is distinguishedby the presence of trabeculae of fibrillary osteoid and woven bone, and psammomatoid JOF is characterised by the presence of small uniform spherical ossicles that resemble psammoma bodies [10]. Psammomatoid JOF is reported more commonly than trabecular JOF [11]. Psammomatoid JOF occurs predominantly in the sinonasal and orbital bones, and trabecular JOF pre- dominantly affects the jaws. Psammomatoid JOF has aggressive behaviour, and it has a very strong tendencyto recur [10,12]. This article presents a case of mandibularJOF in an 8-year-old girl.

3. Case report

An 8-year-old girl was referred to the Maxillofacial Surgery Department with a painless, rapidly enlarging swelling associated with mandibular anterior region of6 months duration. On examination, we noticed the tumor mass was protruding out of the oral cavity and was extending from canine to canine region with expansion of both buccal and lingual cortical plates, theteeth associated with the swelling were displaced and mobile, the mucosa over the lesion was ulcerated because of impingement by the opposing teeth (Fig. 1).The mandibular swelling was firm in consistency and was non-tender.

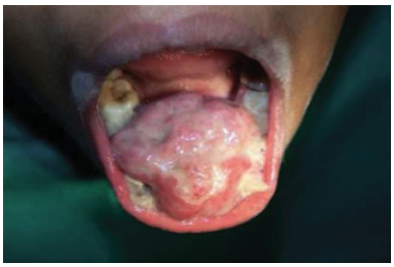

Figure 1: Lesion before biopsy showing expansion cortices and displacement of teeth.

CT scan revealed a well-defined expansile lesion about 5 cm X 3 cm in size extending from canine to canine region; lower border of mandible was intact with normal bone demarcation and central radiopacities (Figs 2 and 3). Biopsy of the lesion was performed, and the histopathology revealed fibroblastic proliferation with new centers of osteoid and numerous multinucleated giant cells, confirming diagnosis of trabecular-type JOF. Within a week after biopsy, there was a flare-up of the lesion, demonstrating its aggressive behaviour.

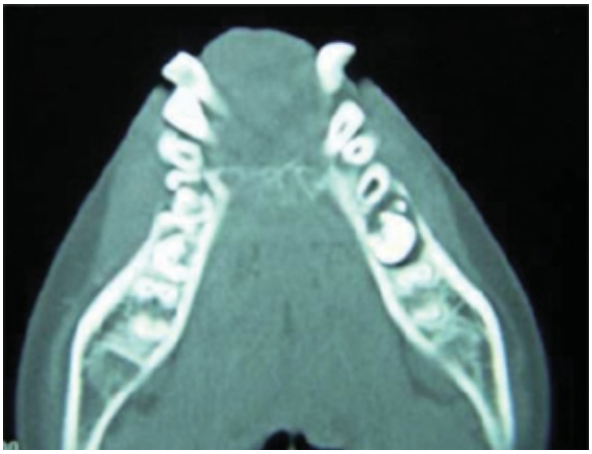

Figure 2: Axial computed tomography showing expansion of corticeswith central radiopacities

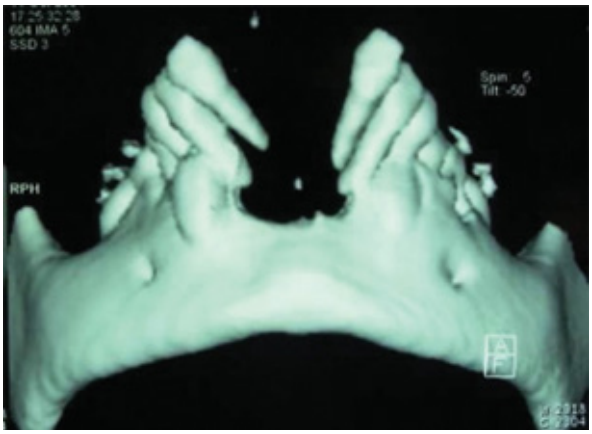

Figure 3: Three-dimensional computed tomography showing well-demarcated lesion with few radiopaque spicules

Complete excision of relatively solid tumor mass was performed intraorally under general anesthesia. After excision of the tumor, the walls of the bone cavity were curetted, and the surgical site was closedprimarily. The healing period was uneventful, and the patient is on a regular follow-up of 2 years with no recurrence (Fig. 4).

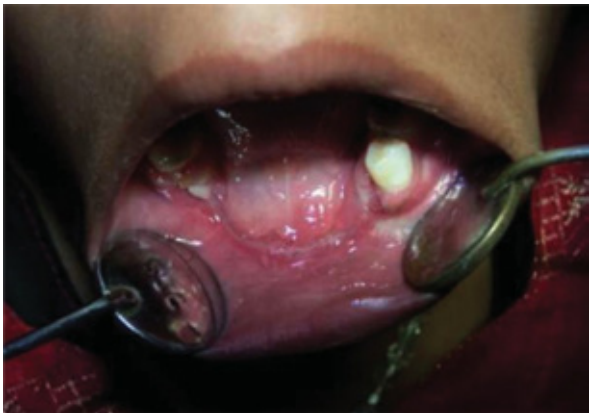

Figure 4: Follow-up after 1 year, showing no signs of recurrence

4. Discussion

Maxillofacial fibro-osseous lesions usually present a diagnostic dilemma for clinicians and pathologists. The JOF is a fibro-osseous lesion that occurs inthe facial bones [6]. It is also called aggressive ossifying fibroma due to its aggressiveness and high tendency to recur, unlike other fibro-osseous lesions, such as cemento-ossifying fibroma, which may resemble radiographically [13]. Due to its distinct histological features, JOF has been recognized as a separate histopathological entity among the fibro-osseous group of lesions [14].

JOF affects both males and females equally withoutany significant gender predilection. However, some researches showed that it is more common among men [15]. In contrast, Johnson et al. [16] stated that mandibular tumors are more frequently common in girls between the age of 5–11 or during the second to fourthdecades of life [3]. The JOF is characterized as expansive, having defined sclerotic borders, locally aggressive and destructive at cortex on CT scan. This lesion is observed as a soft-tissue mass with internal calcification, linear or irregular bone focuses [6,19]. An increase in diffuse contrast is seen after intravenous injection [19]. The radiolucency of the lesion may vary depending on the maturation stage and amount of the calcification [20].

Histologically, the typical features of JOF are a densely cellular stroma containing calcified components. The degree of maturation of the calcification can vary, but the peripheral osteoid rims surrounding the mineral components are an important feature in JOF [21]. The highly cellular nature of the fibrous matrix and woven bone reflects the more aggressive behaviour ofthe tumor. Similar histological features were present in our case, and it was also very aggressive in nature. If JOF does not have adequate surgical treatment, it may have a high rate of recurrence [2,5]. The recurrences are generally seen at an early stage, and they are more aggressive when compared with primary lesions [2].

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There is no consensus on the treatment of JOF cases. Radical resection, local excision conservatively or enucleation with curettage are among the treatment alternatives [2,9]. Slootweg and Müller [7] suggested that there were no differences between the cases that have limited surgical treatment and those with major surgery in terms of results, and they recommended conservative surgery. On the other hand, Waldron [22] suggested that local excision and curettage should be a more preferable method and added that local surgical excision can be applied for recurrent tumor treatment. However, rate of recurrence after conservative treatment was reported in 30–58% of cases [2].

Incomplete resection causes recurrence in aggressive tumors. Therefore, some authors were recommendedenbloc resection as an adequate treatment. Curettage together with peripheral osteotomy or sometimes segmental mandibular resection and mandibular reconstruction are suggested in prevalent or recurrent cases [2]. Sarcomatous degeneration is reported to develop in lesions that have recurrence in long term [13]. In contrast, Espinosa et al. [2] reported a case of unusual bone regeneration after resection of JOF. Secondary mandibular reconstruction with autogenous grafts was delayed dueto the rapid bone formation.

Almost all cases of JOF should be treated surgically. Radiotherapy seems contraindicated because the tumors are believed to be radio-resistant and because of the adverse effects of radiotherapy [23]. The mainstay oftreatment, according to many authors, is curettage of the lesion. For recurrences, accelerated growth and lack of delineation between the lesion and the sur- rounding bone, resection is recommended [4]. Tortorici et al. have also reported a similar case in mandibular premolar region in which they have done a conservative resection with a follow-up of 1 year.In our case, we have reported JOF in the mandibular anterior region which is a less common finding. Histologically, it was of trabecular type, which is less common than the psammomatoid types. The follow-up of this case for 2 years indicates the benefits of conservative surgical treatment. In this case, normal growth, mastication, aesthetics and neural function in the mandible were preserved.

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