CASE REPORT

A large mass in the mandible of an eight year old child

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ABSTRACT

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Facial swellings in children may commonly arise from infection, jaw cysts or benign-malign tumors. Differential diagnosis should be handled considering onset of the symptoms, growth rate and child's age. Fibro-osseous lesions are a group of lesions characterized by fibrosis and contain varying amounts of calcified tissue resembling bone, cement or both. Such lesions may cause enlargement and swelling in the jaws. Juvenile ossifying fibroma (JOF) is one of the fibro-osseous lesions usually seen in children and adolescents. It has an aggressive character with a high recurrence rate. Mandible is more affected than maxilla and psammomatoid type is more common than trabecular type histologically. Bone resection is generally preferred for the treatment due to nature of the tumor.

Here we report a rare case of trabecular type JOF located in the mandible of an 8-year-old child treated by conservative surgery.

KEYWORDS

Conservative surgery, fibro-osseous lesion, juvenil ossifying fibroma, trabecular type

ÖZ

Sekiz yaşındaki çocuğun alt çenesinde geniş bir kitle

Çocukların yüzünde görülen şişlikler daha çok enfeksiyon, çene kistleri ve benign-malign tümörler nedeniyle oluşmaktadır. Ayırıcı tanıda bulguların başlaması, şişliğin gelişme hızı ve çocuğun yaşı göz önüne alınmalıdır. Fibroosseöz lezyonlar fibrosis ile karakterize, kemiğe, semente veya her ikisine benzeyen yapıda değişik oranlarda kalsifiye doku içeren lezyonlardır. Bu tür lezyonlar çenelerde genişlemeye ve şişliğe neden olabilirler. Juvenil ossifiye fibroma (JOF) genelde çocuklarda ve ergenlerde görülen fibroosseöz lezyonlardan biridir. Agresiv karakterde ve yüksek nüks oranına sahiptir. Mandibulada yerleşim maksilladan daha fazladır ve histolojik olarak psammomatoid tipi trabeküler tipinden daha sık görülür. Tümörün yapısı nedeniyle tedavide genellikle çene rezeksiyonu tercih edilir.

Bu olgu sunumunda 8 yaşındaki çocuk hastanın mandibulasında gelişen ve nadir görülen trabeküler tipte JOF ve tedavisi ele alınmaktadır.

ANAHTAR KELİMELER

Konservatif tedavi, fibroosseöz lezyon, jevenil ossifiye fibroma, trabeküler tip,

Juvenile ossifying fibroma (JOF) is a benign fibro-osseous tumor which has aggressive nature. It is mostly diagnosed in the first and second decades and is thought to arise from differentiation of mesenchymal cells of periodontal ligament, multipotent precursor cells, forming into fibrous tissue, cementum or osteoid. Two percent of all oral tumors in children is JOF. Males and females have equal predilection.[1] About 85% of JOF is seen in facial bones. Ratio of occurrence in calvarium and mandible is 12% and 10% respectively.[2] This tumor has a more aggressive character than conventional ossifying fibroma.[3] Trabecular type is mostly located in jaws and has lower aggressive behaviour compared with psammomatoid type. Mandibular lesions of JOF are rarely seen and can be misdiagnosed because of similar radiographic features with odontogenic cysts.[1] The lesion may disrupt the cortical bone and invade into adjacent anatomic structures and soft tissues. Radiopacities can be detected due to variable amount of calcifications and/or linear or irregular focal bone. Computed tomography, magnetic resonance imaging, histopathologic evaluation are beneficial for accurate diagnosis.[4] This report presents a rare case of trabecular type juvenile ossifying fibroma in an 8-year-old child.

CASE

An eight-year-old boy was referred to our clinic with swelling in the right mandibular corpus region. The swelling was first noticed by the patient's parents about 7 months previously. In clinical examination swelling through the buccal, lingual and inferior mandible was noticed (Figure 1). The overlying skin and mucosa were intact. A solid mass was detected and luxation of the involved teeth were observed.

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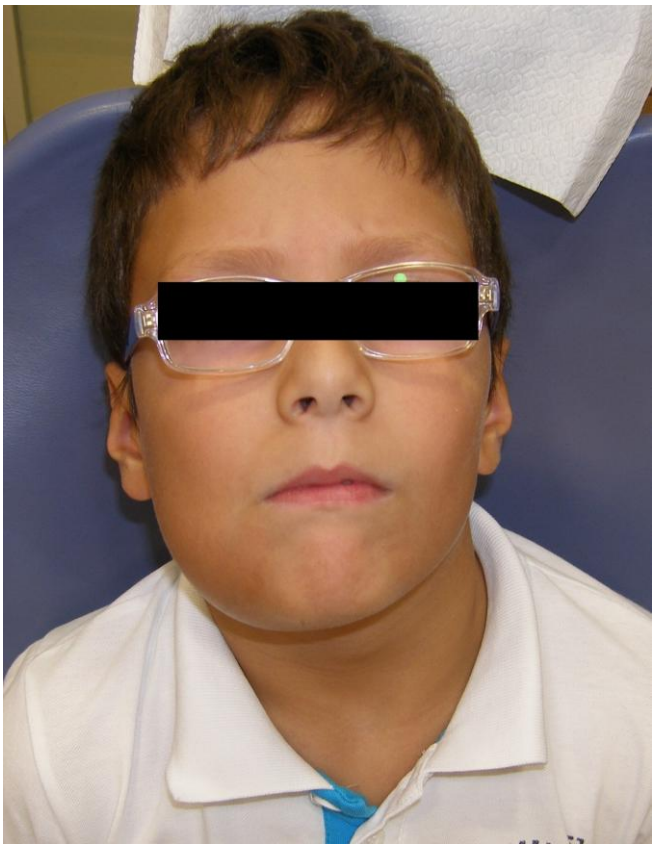

Figure 1.

A swelling through the buccal, lingual and inferior mandible was noticed

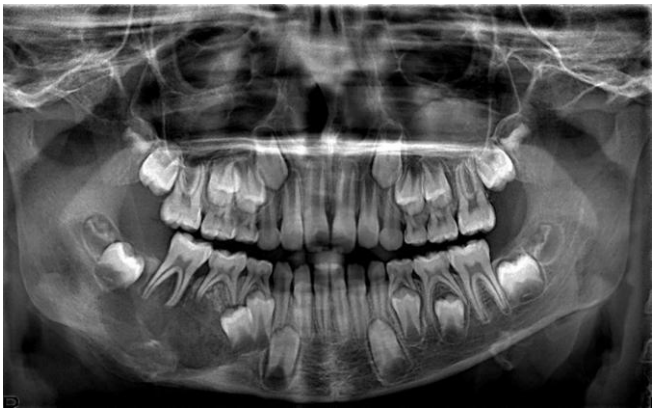

Figure 2.

A large radiolucent lesion in the right posterior mandible

Panoramic radiography revealed a large radiolucent lesion in the posterior mandible with ill-defined borders. The lesion extended from right impacted second premolar to second molar (Figure 2). Axial and sagittal sections of Cone Beam Computerized Tomography (CBCT) presented a large mass containing radioopaque material. The mass enlarged through the buccal, lingual and inferior mandible. While the buccal cortical bone was intact, lingual cortex was perforated and lesion showed slightly soft tissue invasion. (Figure 3) Blood tests yielded no significant information and any signs of Brown Tumor of hyperparathyroidism.

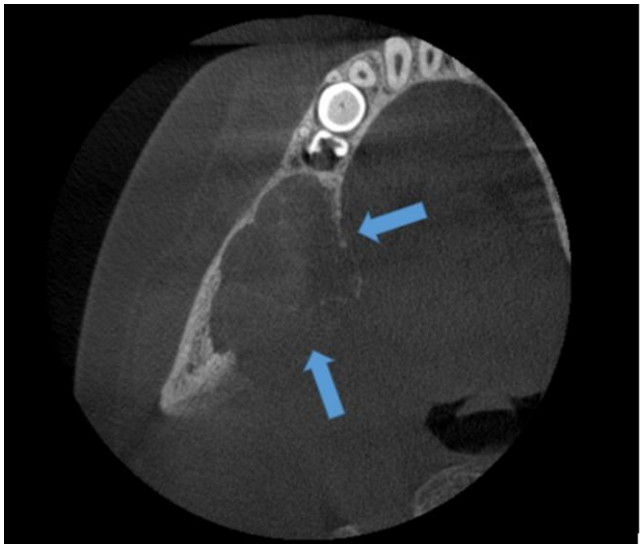

Figure 3.

Axial CBCT image showing lingual soft tissue invasion

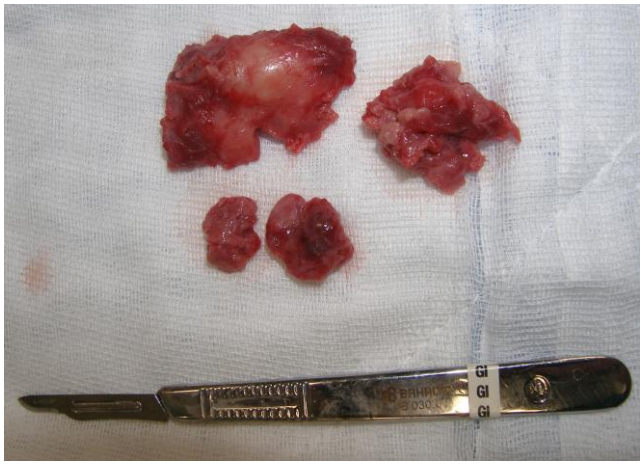

Figure 4.

Excised specimen

Insicional biopsy was performed under local anaesthesia through the extraction socket of first molar and tissue sample was sent for histopathologic evaluation. Provisional diagnosis was juvenile ossifying fibroma. Under general anaesthesia mucoperiosteal flap was elevated and erupted 46#, 85# and 84# teeth and unerupted 45#, 47# were extracted. Lesion was completely removed and surrounding tissue was curetted (Figure 4). Wound was primarily closed with a resorbable suture. Histopathologic examination revealed a cellular tumor in the fibrous stroma. The proliferation of fusiform cells was distinct with woven bone

formation. Multinucleated giant cells and fresh hemorrhage were also observed in the lesion. The mineralized component showed irregular strands of highly cellular sealing osteocytes which were characterized trabecular variant of juvenile ossifying fibroma (Figure 5, Figure 6).

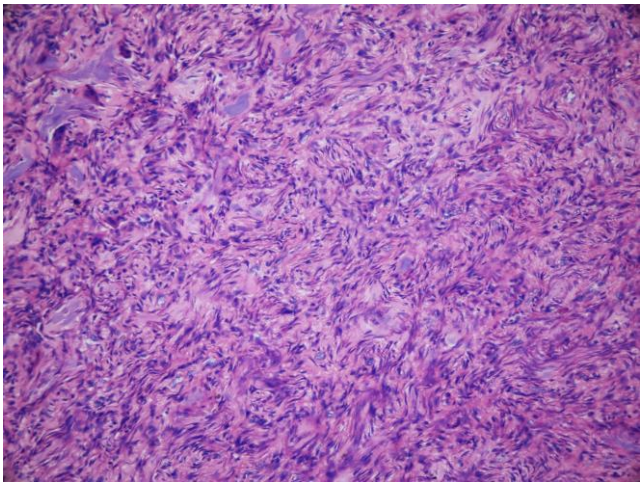

Figure 5.

Cellular proliferation of spindle cells with deposition of osteoid (H&E x400)

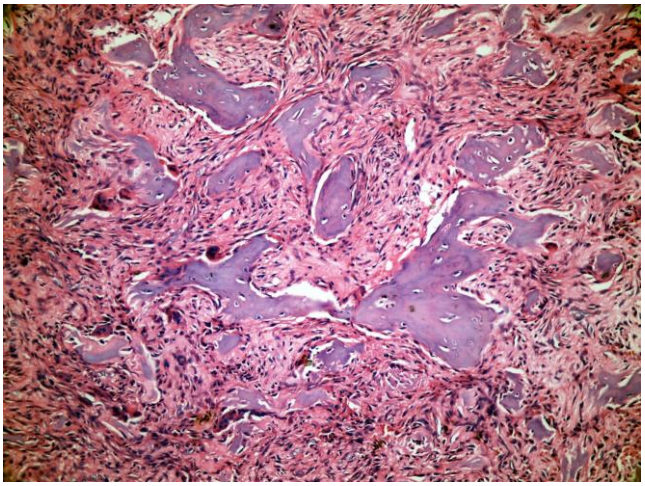

Figure 6.

Osteoid and woven bone surrounded by osteoblasts and osteoclasts (H&E x400)

Although the lesion was completely removed, recurrence can be expected with this type of lesion. Therefore close long-term follow is planned. The patient has been followed-up for every 3 months and follow-up examination at 18 months was uneventful. Panoramic radiography revealed bone healing areas and eruption of 44# tooth (Figure 7). Unerupted third molar associated with the lesion was not extracted at the time of surgery. We expected proximal reposition as the lesion was removed, but control radiograph revealed no improvement in its position (Figure 7).

Unerupted third molar associated with the lesion was not extracted at the time of surgery. We expected proximal reposition as the lesion was removed, but control radiograph revealed no improvement in its position (Figure 7). Therefore further surgery is planned for its removal when the child grows-up.

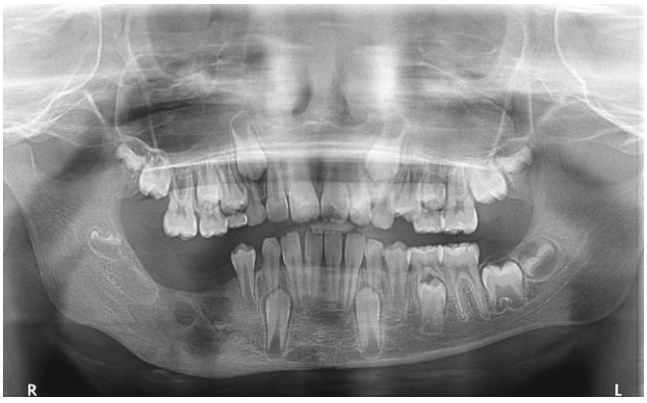

Figure 7.

Post-operative control radiography after 6 months

DISCUSSION

Fibro-osseous lesions are benign jaw lesions in which bone tissue is replaced by fibrous tissue with amorphous mineralization. Juvenile ossifying fibroma is an uncommon tumor which has distinct clinical features. It is a variant of ossifying fibroma (OF) that is generally seen in the cranial and facial bones of young patients. Local aggressive growth is an important clinical feature that distinguishes JOF from OF.[5] JOF occurs as a swelling with pain, whereas OF represents painless swellings in the jaws. OF can occur at any age, mainly between 30-40 years of age, but JOF is encountered in patients younger than 15 years of age. However according to a report by Johnson et al; the age at onset of JOF ranges from 3 months to 72 years.[6] The mean age of trabecular type juvenile ossifying fibroma is reported to be about 11 years. The ages of patients diagnosed with psammomatoid type JOF approaches to 72 years of age. Both types show maxillary predominance and psammomatoid type is more frequent than trabecular type. They are usually discovered with routine radiographs unless clinically detectable facial enlargement is present. Our patient was an 8 –year-old boy who referred to our clinical with a complaint of swelling. Lesions may grow through the neighboring structures and may cause paranasal expansion, facial asymmetry, nasal obstruction, exophthalmos and proptosis.[7, 8] Radiographic appearance of JOF has characteristic features such as; round, well-defined margins similar to cyst-like appearance. Tumors appear as radiolucent, mixed, radiopaque and ground glass-like lesions.

Psammomatoid type usually has ground glass-like features and trabecular type has mixed or radiolucent features. Differential diagnosis of JOF include; fibrous dysplasia, osseous dysplasia, odontoma, and ameloblastoma.[9]

The clinical management and prognosis of juvenile ossifying fibroma are not clear. Conventional OF is treated by surgical excision and recurrence is rare. JOF usually has a slowly growing pattern; however, some demonstrate rapid enlargement. Treatment of juvenile ossifying fibroma depends on aggressiveness of the lesion but not to histological type. Recurrence rate following surgery was reported to be between 30-58%.[6] This rate increases to 67% after conservative surgery and is about 0% after radical surgery. Conservative treatment is preferred when the tumor behaves less aggressive. Conservative treatment includes local excision and curettage to limit facial deformity and dysfunction and thereby to preserve growth and development of the jaws.[10, 11] Conservative treatment also preserves chewing and nerve functions in young patients.[12, 13] Resection may be performed if there is a recurrence, invasion to adjacent site or where the border of mandible is not feasible.[14] Defect reconstruction can be performed at the time of surgery or in a second stage surgery. In our patient we preferred complete removal and curettage since the patient was young and had unerupted teeth. Follow-up at 18 months disclosed no recurrence but close monitoring is necessary since one unerupted tooth in relation to tumour was left in place.

CONCLUSIONS

Facial swellings in children should be handled with caution as these swellings may result from aggressive or malign tumors. Juvenile ossifying fibroma is a benign but aggressive tumor with a high recurrence rate. Although radical surgery is recommended, conservative approach may be preferred considering patient' s age, dentition and localization of the tumor. Close follow-up is mandatory if conservative surgery is performed.

REFERENCES

1. Patil RS, Chakravarthy C, Sunder S, Shekar R. Psammomatoid type juvenile ossifying fibroma of mandible. Ann Maxillofac Surg 2013; 3: 100-3.
2. Antonoglou GN, Sándor GK, Koidou VP, Papageorgiou SN. Non-syndromic and syndromic keratocystic odontogenic tumors: systematic review and meta-analysis of recurrences. J Craniomaxillofac Surg 2014; 42: 364-71.
3. El-Mofty S. Psammomatoid and trabecular juvenile ossifying fibroma of the craniofacial skeleton: two distinct clinicopathologic entities. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2002; 93: 296-304.
4. Solomon M, S Khandelwal, A Raghu, S Carnelio. Psammomatoid Juvenile Ossifying Fibroma of the Mandible – A Histochemical insight!. The Internet Journal of Dental Science. 2008 Volume 7 Number 2.
5. Phattarataratip E, Pholjaroen C, Tiranon P. A Clinicopathologic Analysis of 207 Cases of Benign Fibro-Osseous Lesions of the Jaws. Int J Surg Pathol 2014; 22: 326-33
6. 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.
7. Shields JA, Peyster RG, Handler SD, Augsburger JJ, Kapustiak J. Massive juvenile ossifying fibroma of maxillary sinus with orbital involvement. Br J Ophthalmol 1985; 69: 392-5.
8. Guttikonda V, Taneeru S, Gaddipati R, Madala J. A massive sinonasal psammomatoid variant of juvenile ossifying fibroma: Report of a rare entity. J Oral Maxillofac Pathol 2013; 17: 302-5.
9. Neville BW, Damn DD, Allen CM, Bouquot JE. Oral and Maxillofacial Pathology, 2 nd ed. Philadelphia, WB Saunders;. 2002; p. 564-5.
10. Slootweg PJ, Panders AK, Koopmans R, Nikkels PGJ. Juvenile ossifying fibroma. An analysis of 33 cases with emphasis on histopathological aspects. J Oral Pathol Med 1994; 23: 385-8.
11. Papadaki ME, Troulis MJ, Kaban LB. Advances in diagnosis and management of fibro-osseous lesions. Oral Maxillofac Surg Clin North Am 2005; 17: 415-34.
12. Abuzinada S, Alyamani A. Management of juvenile ossifying fibroma in the maxilla and mandible. J Maxillofac Oral Surg 2010; 9: 91-5.
13. Han J, Hu L, Zhang C, Yang X, Tian Z, Wang Y, et al. Juvenile ossifying fibroma of the jaw: a retrospective study of 15 cases. Int J Oral Maxillofac Surg 2016; 45: 368-76.
14. Smith SF, Newman L, Walker DM, Papadopoulos H Juvenile aggressive psammomatoid ossifying fibroma: an interesting, challenging, and unusual case report and review of the literature. J Oral Maxillofac Surg. 2009; 67: 200-6.

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