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

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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 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. 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.

### ABSTRACT

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Facial cysts 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, juvenile ossifying fibroma, trabecular type

### ÖZ

Sekiz yaşındaki çocuğun alt çenesinde geniş bir kitle


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

### ANAHTAR KELİMELER

Konservatif tedavi, fibroosseöz lezyon, juvenil ossifiye fibroma, trabeküler tıp,
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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 radiopaque 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.

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).

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). Therefore further surgery is planned for its removal when the child grows up.

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 clinic 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 benign 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


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