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Juvenile ossifying fibroma of the jaw: a retrospective study of 15 cases

J. Han[1,5], L. Hu[1,5], C. Zhang[2], X. Yang[1], Z. Tian[2], Y. Wang[1], L. Zhu[3], C. Yang[4], J. Sun[1], C. Zhang[1], J. Li[2], L. Xu[1]

[1]Department of Oral and Maxillofacial–Head and Neck Oncology, Ninth People's Hospital, Shanghai Jiao Tong University School of Medicine, Shanghai, China; [2]Department of Oral Pathology, Ninth People's Hospital, Shanghai Jiao Tong University School of Medicine, Shanghai, China; [3]Department of Radiology, Ninth People's Hospital, Shanghai Jiao Tong University School of Medicine, Shanghai, China; [4]Department of Oral Surgery, Ninth People's Hospital, Shanghai Jiao Tong University School of Medicine, Shanghai, China

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Abstract. The management of patients with juvenile ossifying fibroma (JOF) remains controversial. To explore the correlations between different treatments and the patient prognosis, 15 cases of JOF of the jaw were reviewed. Five patients were male and 10 were female. Patient age at the time of disease onset ranged from 7 to 18 years (mean 10.9 years). Nine tumours were located in mandible and six in the maxilla. These cases typically manifested clinically as painless swelling of the jaw (9/15, 60%); 40% (6/15) of the cases were associated with pain, diplopia, stuffy nose, and/or rapid growth. Images of JOF can show a radiolucent, mixed, or ground glass-like appearance. Pathological examinations revealed 10 cases of juvenile trabecular ossifying fibroma (JTOF) and five cases of juvenile psammomatoid ossifying fibroma (JPOF). In terms of the treatment plan, six patients initially received radical surgery; nine patients underwent conservative treatment, among whom six (6/9, 66.7%) had one or more recurrence. At the end of the follow-up period, 12 patients had no evidence of tumour recurrence and three cases were alive with a tumour. In summary, surgeons should develop the surgical plan according to the extent of the lesion, relapse status, growth rate, and family choice, and these patients should be followed up closely.

Key words: jaw; JOF; JPOF; JTOF; treatment.

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Ossifying fibroma (OF) is a type of benign fibro-osseous lesion. This tumour is characterized by clear boundaries and cell-rich fibrosis, and contains varying amounts of calcified tissue resembling bone, cementum, or both. OF lesions are classified as conventional ossifying fibroma and juvenile ossifying fibroma (JOF).[1] Conventional OF is mainly seen in adults. JOF usually occurs in children or adolescents, and it is therefore also known as juvenile active/aggressive ossifying fibroma (JAOF). In 2005, the World Health Organization (WHO) noted that the age at onset of JOF is 15 years and younger.[1] However, JOF has also been documented in adults.[2,3]

JOF has characteristics of aggressive growth, is associated with damage to the cortical bone, and may involve the nasal cavity, eyes, and even cerebrum. However, there are no reports of cases of malignant transformation or metastasis. The imaging characteristics of JOF show expansive, well-defined radiolucent or mixed images that are separated from the surrounding normal bone.[4] Based on histopathology, JOF is divided into the

[5] These authors contributed equally to this work.

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following two subtypes: juvenile psammomatoid ossifying fibroma (JPOF) and juvenile trabecular ossifying fibroma (JTOF). In addition, JOF usually contains multinucleated giant cells, whereas conventional OF does not.[5,6] Conventional OF usually presents as a slow-growing mass of bone expansion that is usually without symptoms and rarely recurs; conversely, 38.5% of JOF cases are associated with mandibular swelling and pain and there may be short-term rapid growth.[7] Therefore, surgeons should consider clinical features, radiological characteristics, and pathological features when diagnosing JOF. Key points to be considered include age, status of growth, and histopathology.[8]

The management of patients with JOF remains controversial. According to the literature, the recurrence rate after surgery is approximately 30–58%.[1,3,9–11] Therefore, designing an appropriate treatment plan for the JOF patient could help to improve the prognosis and quality of life. Depending on biological behaviour and the lesions involved, treatment can be conservative or radical.[12] Some researchers have emphasized the aggressive growth characteristics and high recurrence rate of JOF, leading them to advocate local radical surgery.[4,13,14] In contrast, others consider that conservative treatment is more beneficial for young patients when taking into account their growth and development, their appearance, and the preservation of chewing and nerve functions.[12,15,16]

This study was performed to review the clinical features, imaging features, and pathological characteristics of 15 cases of JOF treated at the authors' institution. The relationships between the different treatment options and the prognosis are discussed and the most suitable treatments for JOF are explored.

Materials and methods

A total of 15 patients with JOF treated between 2005 and 2014 in the Department of Oral and Maxillofacial–Head and Neck Oncology of the study institution were included. Each patient's medical history was reviewed carefully to collect clinical data (including age, sex, disease location, symptoms, surgical approach, and prognosis), radiological features, histological type, and other information.

According to the anatomical location, the mandible was divided into anterior (left canine to right canine), posterior (first premolar to third molar), angle, ramus, and condyle. The maxilla was divided into anterior (left canine to right canine) and posterior (first premolar to the maxillary tuberosity). Imaging data were used to analyze the lesion and surrounding tissue boundaries and the internal radiographic architecture. The internal radiographic architecture was divided into mixed radiopaque and radiolucent, unilocular or multilocular radiolucency, and ground glass opaque. An oral pathology specialist classified the histological subtype as either JTOF or JPOF based on the 2005 WHO classification criteria for head and neck cancer.[1] Surgeries performed included conservative treatments (curettage and

Table 1. Clinical information, treatment, and follow-up information for the 15 cases of juvenile ossifying fibroma (JOF).

Patient No./sex	Age at onset (years)	Age at operation (years)	Size	Symptoms	Location	Surgery	Follow-up (months); outcome
Conservative treatment							
1/M	8	8	4.5 × 4	Swelling	Right maxilla	Curettage	72; alive with tumour
2/F	14	19	2 × 1.5	Swelling	Left maxilla (posterior)	Curettage	20; alive with tumour
3/M	13	13	5 × 4	Swelling	Right mandible (posterior)	Enucleation	38; NED
4(R)/M	18	20	3 × 2	Swelling, pain	Left maxilla	Curettage	124; alive with tumour
5(R)/F	9	16	6 × 4	Swelling	Right mandible (posterior)	Curettage/resection + fibula	95; NED
6(R)/F	12	15	5 × 4	Swelling	Left mandible (posterior/angle)	Curettage/resection + iliac crest	146; NED
7(R)/M	15	17	9 × 8	Swelling, rapid growth	Right mandible (posterior)	Curettage/resection + fibula	105; NED
8(R)/M	10	12	10 × 7	Swelling, rapid growth	Left mandible	Curettage/resection + fibula	40; NED
9(R)/F	9	19	9 × 8	Swelling, stuffy nose, diplopia	Right maxilla	Curettage/resection + fibula	128; NED
Radical treatment							
10/F	13	19	12 × 8	Swelling	Left mandible (posterior)	Resection + fibula	87; NED
11/F	11	11	6 × 4	Swelling	Right mandible (posterior)	Resection + iliac crest	58; NED
12/F	7	7	6 × 4.5	Swelling	Left mandible (posterior/angle)	Resection + iliac crest	36; NED
13/F	7	7	3 × 2	Swelling	Right mandible (condyle)	Resection + costal cartilage	15; NED
14/F	7	8	4 × 3.5	Swelling, pain, diplopia, rapid growth	Left maxilla (posterior)	Resection	18; NED
15/F	11	15	6 × 4	Swelling, pain, rapid growth	Right maxilla	Resection	13; NED

F, female; M, male; NED, no evidence of disease; (R), recurrence.