СЛУЧАЙ ИЗ ПРАКТИКИ

DISTINCT VARIANT OF JUVENILE OSSIFYING FIBROMA OF THE MANDIBULAR RAMUS

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Purpose. This case report documents a rare instance of juvenile trabecular ossifying fibroma (JTOF) in the mandibular ramus, an unusual location with only one prior reported case. The study aims to expand knowledge of JTOF's anatomical distribution and highlight the importance of comprehensive diagnostic evaluations for pediatric maxillofacial lesions.

Materials and Methods. A 10 year-old female presented with facial pain and swelling. Diagnostic tools included panoramic radiography and cone beam computed tomography (CBCT), followed by an incisional biopsy for histopathological confirmation. Treatment involved surgical resection of the affected mandible and reconstruction using an iliac bone crest graft, with post-operative monitoring for healing and recurrence.

Results. Imaging revealed extensive erosion and radiolucencies in the right mandibular ramus. Histopathology confirmed JTOF. Post-surgery, the patient showed satisfactory healing, with no complications and good bone graft integration. Facial asymmetry resolved, and mandibular function was restored.

Discussion. JTOF typically affects the maxilla but rarely the mandibular ramus. The aggressive nature of JTOF necessitates accurate imaging and histopathological analysis for diagnosis. Differential diagnoses include fibrous dysplasia and osteosarcoma. The study emphasizes the need for thorough evaluation and surgical intervention to manage such lesions effectively.

Conclusion. This case highlights the rarity of JTOF in the mandibular ramus, contributing to clinical knowledge. It underscores the importance of comprehensive radiological and histopathological assessments in pediatric jaw lesions. Clinicians should consider JTOF in differential diagnoses to ensure timely and appropriate treatment, optimizing patient outcomes.

Keywords: Ossifying Fibroma, Mandibular Neoplasms, Mandibular Ramus, Bone Neoplasms, Adolescent.

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РЕДКИЙ ВАРИАНТ ЮВЕНИЛЬНОЙ ОССИФИЦИРУЮЩЕЙ ФИБРОМЫ ВЕТВИ НИЖНЕЙ ЧЕЛЮСТИ

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Цель исследования. Данное клиническое наблюдение описывает редкий случай ювенильной трабекулярной оссифицирующей фибромы (ЮТОФ) в области ветви нижней челюсти, что является необычной локализацией, ранее был зарегистрирован только один подобный случай. Исследование направлено на расширение знаний об анатомическом распределении ЮТОФ и подчеркивает важность комплексной диагностической оценки при педиатрических поражениях челюстно-лицевой области.

Материалы и методы. Девочка 10-ти лет обратилась с жалобами на боль и припухлость в области нижней челюсти. Для диагностики использовались панорамная рентгенография и конусно-лучевая компьютерная томография (КЛКТ), после чего была проведена инцизионная биопсия для гистопатологического подтверждения. Лечение включало хирургическую резекцию пораженного участка нижней челюсти и реконструкцию с использованием трансплантата из гребня подвздошной кости. После операции проводилось наблюдение за заживлением и возможным рецидивом.

Результаты. Визуализировались обширная эрозия и рентгенопрозрачные участки в области правой ветви нижней челюсти. Гистопатологическое исследование подтвердило диагноз ЮТОФ. После операции отмечено удовлетворительное заживление, без осложнений, хорошая интеграция костного трансплантата. Асимметрия лица была устранена, функция нижней челюсти восстановлена.

Обсуждение. Ювенильная трабекулярная оссифицирующая фиброма обычно поражает верхнюю челюсть, но крайне редко встречается в области ветви нижней челюсти. Агрессивный характер ЮТОФ требует точной визуализации и гистопатологического анализа для постановки диагноза. Дифференциальная диагностика включает фиброзную дисплазию и остеосаркому. Исследование подчеркивает необходимость тщательной оценки и хирургического вмешательства для эффективного лечения подобных поражений.

Заключение. Данный случай подчеркивает редкость ювенильной трабекулярной оссифицирующей фибромы в области ветви нижней челюсти, внося вклад в клинические знания. Он акцентирует важность комплексной радиологической и гистопатологической оценки при педиатрических поражениях челюстей. Клиницисты должны учитывать ЮТОФ при дифференциальной диагностике, чтобы обеспечить своевременное и адекватное лечение, оптимизируя результаты для пациентов.

Ключевые слова: оссифицирующая фиброма, новообразования нижней челюсти, ветвь нижней челюсти, костные новообразования, подростки.

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The maxillofacial region in paediatric patients, particularly during the mixed dentition phase, is susceptible to significant developmental perturbations due to neoplastic and tumour-like lesions, potentially leading to substantial morbidity. These lesions may originate from either odontogenic or non-odontogenic tissues. The prevalence of dental anomalies in the paediatric population is often attributed to inflammatory, infectious, and reactive processes, which can exhibit radiographic characteristics mimicking more aggressive locally invasive neoplasms or malignant conditions [1]. Consequently, invasive histopathological evaluation is frequently necessitated for definitive diagnosis and appropriate management.

The radiographic presentation of osseous lesions in the juvenile mandible and maxilla, including their dimensions, morphology, extent of osseous involvement, and impact on developing permanent tooth germs, is of paramount diagnostic importance [2]. While panoramic radiography is commonly employed as an initial diagnostic tool, its limitations, such as two-dimensional representation resulting in structural superimposition, are well recognised. Therefore, advanced imaging modalities, particularly cone beam computed tomography (CBCT) with its superior spatial resolution, are indispensable for accurate delineation and characterisation of pathological entities.

This case report elucidates an exceptionally rare manifestation of trabecular-type juvenile ossifying fibroma affecting the mandibular ramus in a middle childhood female patient. The discussion encompasses the clinical presentation, diagnostic challenges encountered, and the therapeutic approach employed. The case serves to highlight the importance of comprehensive radiological assessment and histopathological correlation in the management of such uncommon paediatric maxillofacial lesions.

Material and methods.

Clinical Description

A 10 year-old female presented with a one-month history of facial pain and swelling on the right side. The patient reported antecedent trauma during play, followed by progressive swelling. She experienced intermittent pain exacerbated by mastication. The patient's medical history was unremarkable, with no systemic illnesses or drug allergies.

Diagnostic Assessment.

Extraoral examination revealed significant facial asymmetry on the right side. A diffuse swelling extended from 0.5 cm inferior to the right infraorbital margin to the inferior border of the mandible superoinferiorly and from 0.5 cm lateral to the right oral commissure to the angle of the mandible anteroposteriorly (fig. 1A, 1B). On palpation, the swelling exhibited soft to firm consistency and was compressible. The right submandibular lymph node was palpable and tender. Intraoral examination revealed no vestibular obliteration or abnormalities in the right posterior dentition (fig. 1C).

Results.

The patient presented with an external orthopantomogram. Panoramic imaging demonstrated diffuse, multiple, ill-defined radiolucencies extending from the mandibular right first

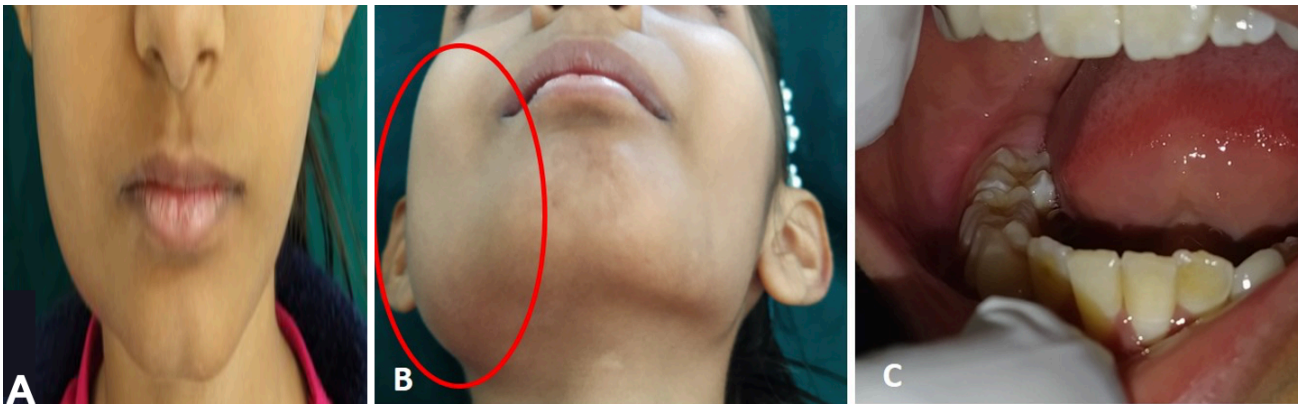

Fig. 1 (Рис. 1)

Fig. 1. Photos.

A – Frontal view showing swelling on right side of the face. B – Inferior view of the swelling. C – Shows right posterior mandibular region.

Рис. 1. Фотографии.

А – Вид спереди, демонстрирующий припухлость на правой стороне лица. В – Вид снизу, показывающий припухлость. С – Область задней части нижней челюсти справа.

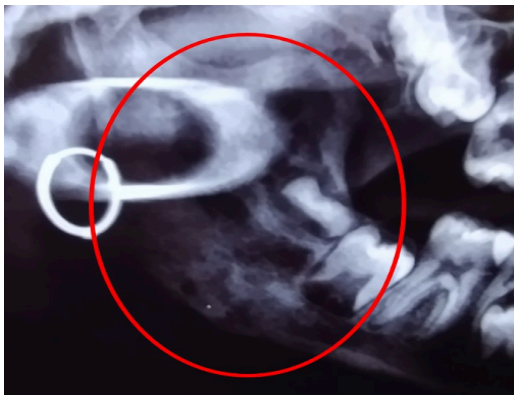

Fig. 2 (Рис. 2)

Fig. 2. Panoramic X-ray.

Cropped panoramic image of right posterior region showing diffuse multiple radiolucency in ramus of the mandible.

Рис. 2. Ортопантомография.

Прицельное панорамное изображение области ветви нижней челюсти справа, демонстрирующее диффузные множественные рентгенопрозрачные участки.

molar, involving the ramus and coronoid processes. A superimposed ghost image obscured the pathology in the ramus region (fig. 2). Consequently, cone beam computed tomography was recommended for further evaluation.

CBCT 3D reconstruction revealed extensive erosion in the right mandibular ramus, sparing the odontogenic region (fig. 3A). Coronal and sagittal views demonstrated diffuse multiple radiolucencies interspersed with radiopaque specks in the right mandible, extending from distal to the first mandibular molar to the ramus, including the coronoid process. Figures 3B and 3D illustrate cortical plate thinning. An axial CBCT view revealed periosteal bone formation in the ramus region (fig. 3C).

Based on the clinical and radiological presentation, juvenile ossifying fibroma was considered the primary diagnosis. The differential diagnosis included fibrous dysplasia, osteoblastoma, osteosarcoma, and cementoblastoma.

An incisional biopsy was performed under local anaesthesia. Histopathological examination revealed spindle-shaped fibroblasts interrupted by irregular strands of woven bone with osteoblastic rimming. Focal areas demonstrated a few multinucleated giant cells. These features were

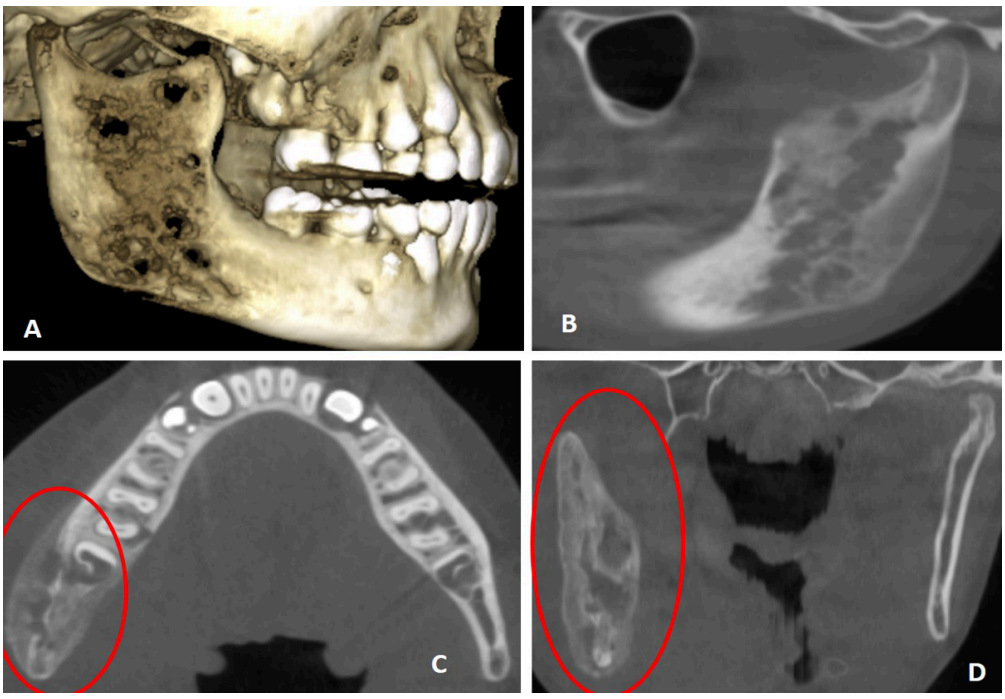

Fig. 3 (Рис. 3)

Fig. 3. CBCT.

A – 3D reconstruction view showing extensive erosion of the right ramus. B, C and D – sagittal, axial, and coronal views showing multiple radiolucency with thinning of the cortical plate and periosteal reaction, respectively.

Рис. 3. КЛКТ.

А – 3D-реконструкция, обширная эрозия правой ветви нижней челюсти. B, C и D – Сагиттальный, аксиальный и коронарный срезы соответственно, множественные рентгенопрозрачные участки с истончением кортикальной пластинки и периостальной реакцией.

consistent with the trabecular variant of juvenile ossifying fibroma.

Differential Diagnosis.

The various differential diagnoses considered in this case have been enumerated in Table №1.

Treatment.

The surgical management of this juvenile ossifying fibroma case was performed under general anaesthesia, administered by a paediatric anaesthesiologist following standard protocols. The procedure involved careful resection of the affected right mandible with attention to preserving vital structures, particularly the facial nerve branches and blood vessels. The reconstruction utilised an iliac bone crest graft, chosen for its high-quality cortical and cancellous bone properties, excellent osteogenic potential, and sufficient bone volume necessary for paediatric mandibular reconstruction.

The harvested iliac crest graft was precisely shaped to match the resected mandibular segment and secured using age-appropriate fixation methods that accommodate future growth. Post-operative care encompassed paediatric pain management, wound care at both surgical sites, and a progressive return to normal feeding. Key elements included long-term monitoring of bone healing, facial growth, dental occlusion, and vigilance for potential recurrence, as the treatment aimed to achieve complete pathological removal while ensuring proper mandibular development throughout the child's growth period.

Outcome and follow-up.

The patient underwent successful surgical excision and reconstruction of the right mandible under general anaesthesia using an iliac bone crest graft. Post-operative healing was uneventful with no immediate complications. Regular follow-up visits were scheduled at 1 week, 1 month, 3 months, 6 months, and annually thereafter. The 1-week follow-up showed satisfactory healing of both surgical sites with minimal postoperative edoema. At 3 months, radiographic examination revealed good integration of the bone graft with no signs of recurrence or complications. The 6-month follow-up demonstrated complete resolution of facial asymmetry with restored mandibular contour and function. Long-term monitoring will continue throughout the growth period to ensure normal facial development and early detection of any potential recurrence.

Discussion.

Ossifying fibroma (OF) is a benign neoplasm characterised by encapsulation and variable quantities of osseous or cementum-like tissues within a fibrous stroma. These fibro-osseous lesions demonstrate a predilection for the gnathic bones, particularly the mandible. OFs are categorised into two distinct subtypes: conventional and juvenile. The conventional variant exhibits indolent growth and clear demarcation from the surrounding osseous matrix. It is also referred to as ossifying fibroma, cemento-ossifying fibroma, or cementifying fibroma and typically manifests in the adult population. In contrast, juvenile OF represents a rare fibro-osseous neoplasm with a lower incidence and more aggressive biological behaviour compared to its conventional counterpart [3].

Juvenile ossifying fibroma (JOF), also termed juvenile active/aggressive ossifying fibroma (JAOF), predominantly affects the paediatric and adolescent demographic. The World Health Organisation (WHO) in 2005 noted that the typical age of onset for JOF is 15 years or younger [4]. JOF is further subdivided into two histological variants: juvenile psammomatoid ossifying fibroma (JPOF) and juvenile trabecular ossifying fibroma (JTOF). While both entities are relatively uncommon, JPOF demonstrates a higher prevalence than JTOF. The case in question presents a rare manifestation of JTOF.

The peak incidence of TJOF occurs between 8 and 12 years, concordant with the present case at 10 years of age, whereas PJOF typically presents between 16 and 33 years. A distinguishing clinical feature between TJOF and PJOF is their anatomical predilection. PJOF predominantly affects the paranasal sinuses, while TJOF exhibits a predilection for the maxilla [5]. This case report documents an atypical presentation of TJOF in the mandibular ramus, thus representing a rare entity. JTOF is typically asymptomatic, with early lesions often discovered as incidental radiographic findings. Dental displacement may serve as an early indicator of the tumour's aggressive growth potential, potentially leading to facial asymmetry and gnathic deformity. Similarly, in the present case, the lesion demonstrated aggressive behaviour resulting in facial asymmetry.

Radiographically, JTOF typically presents as unilateral, unilocular mixed radiolucent/radiopaque lesions. However, they may occasionally appear entirely radiolucent with delicate internal radiopacities. JTOF characteristically demonstrates concentric expansion from a central epicentre, extending centrifugally. This expansion may result in dental displacement and inferior alveolar nerve canal deviation. Notably, the outer cortical plate remains intact, albeit expanded and attenuated. Root resorption is a frequent finding. The neoplasm exhibits rapid progression and may mimic malignant entities. In younger patients, it may be mistaken for osteosarcoma. However, osteosarcomas can be differentiated radiographically by their irregular cortical margins that invade the periodontal ligament and soft tissues [6]. They also lack the thin radiolucent

Table №1. Differential diagnoses.

Clinical Feature	Juvenile Ossifying Fibroma	Aneurysmal Bone Cyst	Traumatic Bone Cyst	Central Giant Cell Granuloma	Fibrous Dysplasia
Age and Gender	Child (Early and middle childhood) Female predominance	Adolescents (First Decade), Female predominance	Adolescents (First Decade), No gender predilection	Young adults (Second and Third Decade), Female predominance	Children/adolescents (Childhood and Adolescence), No gender predilection
Location	Right facial region, Mandibular involvement	Any jaw location, Mandible preferred	Posterior mandible common	Usually anterior mandible	Craniofacial bones, Usually unilateral
Pain	Present, Exacerbated by mastication	May cause significant pain	Usually asymptomatic	May be painful	Usually painless
Swelling	Rapid progressive swelling	Rapid expansion	Minimal expansion	Slow expansion	Gradual expansion
Consistency	Soft to firm, Compressible	Soft, Compressible	Bony hard	Firm to hard	Firm to hard
History of Trauma	Present in this case	May be associated	Common precipitating factor	Not typically associated	Not typically associated
Lymph Node Involvement	Present, Tender	Not typically involved	Not typically involved	Not typically involved	Not typically involved
Growth Pattern	Progressive growth	Progressive expansion	Stabilizes after healing	Can be aggressive locally	Stabilizes after puberty
Additional Features	No vestibular obliteration, Progressive nature	Blood-filled spaces, Shell-like expansion	Usually asymptomatic, Self-limiting	May cause tooth displacement, Root resorption	Associated with bone remodeling, Ground-glass appearance
Match Score	89%	72%	67%	61%	56%

corticated boundary characteristic of JTOF, which is observed in the present case. Contrary to established epidemiological data, this report documents a case of JTOF in the mandibular region, a presentation infrequently reported in the extant literature.

The histopathological criteria for juvenile trabecular ossifying fibroma (JTOF) are characterised by a neoplasm predominantly composed of cellular fibroblastic stroma interspersed with delicate trabeculae of immature bone. These trabecular structures may anastomose, forming a lattice-like configuration. While typically well-demarcated, JTOF lacks encapsulation. Notable variability in stromal cellularity may be observed. A hallmark feature evident in the present case is the presence of prominent osteoblastic rimming surrounding the bony trabeculae.

The histomorphological spectrum of JTOF may encompass foci of osteoclastic giant cells, areas of haemorrhage, and regions of pseudocystic stromal degeneration. Mitotic activity may be appreciated in areas of heightened cellularity [6].

The diagnostic algorithm for JOFs necessitates differentiation from entities such as cementoblastoma, osteoblastoma, and osteosarcoma through a comprehensive evaluation of clinical, radiographic, and histopathological parameters. Vascular neoplasms, particularly central haemangiomas, which exhibit a predilection for the paediatric and young adult population, should also be included in the differential diagnosis. These lesions often present with rapid expansion and may be associated with auscultatory findings such as thrills or bruits [7].

The definitive diagnosis of JTOF relies on the integration of clinical findings with radiographic features, as JTOF shares histomorphological characteristics and overlapping attributes with other fibro-osseous lesions. It is noteworthy that immunohistochemical analysis offers limited utility in differentiating among benign fibro-osseous entities [6].

The prognostic outlook for JTOF is characterised by unpredictability, with a significant propensity for recurrence following surgical intervention. Reported recurrence rates range from 30% to 67%, particularly in cases of incomplete excision. It is imperative to note that JTOF does not possess malignant potential [6].

Conclusion.

In summation, this case presentation of trabecular-type juvenile ossifying fibroma underscores the paramount importance of accurate diagnostic imaging, specifically through cone-beam computed tomography, which proves indispensable in delineating intricate anatomical nuances. While the typical locus of manifestation for juvenile trabecular ossifying fibroma is within the maxillary bone, this particular case exhibits an atypical presentation in the mandibular ramus, thus accentuating the necessity for clinicians to remain cognisant of such anomalous sites of occurrence.

The case in question serves to elucidate the critical nature of including JTOF in the differential diagnostic considerations for osseous lesions of the jaw in the paediatric population. This inclusion is pivotal for facilitating expeditious therapeutic intervention and optimising prognostic outcomes.

The rarity of this presentation, coupled with its distinctive anatomical location, not only contributes to the existing body of literature but also reinforces the imperative for comprehensive radiological evaluation in cases of suspected fibro-osseous lesions in young patients. Furthermore, it highlights the potential for JTOF to manifest in diverse anatomical sites within the craniofacial skeleton, thereby expanding our understanding of its clinical spectrum.

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