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Case Report



Osteoid osteoma of the trapezium bone in a child: A clinical case

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ABSTRACT

BACKGROUND: Osteoid osteoma is a benign neoplasm of bones with a diameter of up to 1.5 cm. It is most common in children in the second decade of life and at the age of 20–30 years, predominantly in men. In large neoplasms (>1.5 cm) with a histological study identical to osteoid osteoma, the pathological process is regarded as osteoblastoma or giant osteoid osteoma. The tumor is most often located in long tubular bones and on the hand in 5%–15% of the patients. Differential diagnosis is carried out with rheumatoid arthritis, inflammation, and other neoplasms. Various options for the treatment of osteoid osteoma, both conservative and operative, have been proposed.

CLINICAL CASE: A 17-year-old patient underwent surgical treatment for trapezium bone neoplasm and pronounced chronic pain syndrome.

DISCUSSION: The identification and treatment of osteoid osteoma remain challenging, which is associated with the similarity of clinical symptoms and ineffectiveness (in the initial stages of its development) of X-ray examination. Thus, computed tomography is necessary to identify a pathological structure. If a formation on the hand is detected, surgery (tumor resection) is advised; if necessary, bone grafting of the defect is performed. After surgical treatment, relapse may occur within 7 months, which is associated with incomplete tumor removal. To confirm the diagnosis, a pathomorphological study is necessary.

CONCLUSIONS: Chronic pain syndrome requires a thorough examination of the patient and a differential diagnosis. Surgical treatment allows the restoration of hand functions and alleviates the tumor-associated pain syndrome.

Keywords: osteoid osteoma; tumor; children; hand surgery.

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Клинический случай

Остеоид-остеома кости-трапеции у ребенка (клиническое наблюдение)

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АННОТАЦИЯ

Обоснование. Остеоид-остеома — доброкачественное новообразование скелета диаметром до 1,5 см, которое наиболее часто встречается у детей на втором десятилетии жизни и в возрасте 20–30 лет преимущественно у лиц мужского пола. Интересно, что при больших размерах новообразования (более 1,5 см) с гистологической картиной, идентичной остеоид-остеоме, патологический процесс расценивают как остеобластому или гигантскую остеоид-остеому. Наиболее часто опухоль выявляют в длинных трубчатых костях и только у 5–15 % пациентов — в костях кисти. На этапе диагностического поиска наиболее часто опухоль приходится дифференцировать с воспалительными костно-деструктивными и ревматоидными процессами. Предложены различные варианты лечения остеоид-остеомы — как консервативные, так и оперативные.

Клиническое наблюдение. Пациенту 17 лет проведено оперативное лечение в связи с патологическим очагом в кости-трапеции и выраженным длительным некупируемым болевым синдромом.

Обсуждение. Выявление остеоид-остеомы (при отсутствии соответствующего опыта) остается сложной проблемой, что связано с неспецифичностью клинических проявлений заболевания и нередко малой информативностью стандартного рентгенологического исследования. Необходимо проведение компьютерной томографии для обнаружения патологического очага. При выявлении опухоли в костях кисти целесообразно выполнять хирургическое вмешательство в объеме резекции патологического очага в пределах неизменной костной ткани и при необходимости осуществлять костную пластику пострезекционного дефекта. При нерадикальном удалении опухоли характерный болевой синдром возобновляется в ближайшие сроки после оперативного вмешательства. Для подтверждения клинического диагноза необходимо проведение патоморфологического исследования.

Заключение. При длительном некупируемом болевом синдроме необходимо комплексное обследование пациента. Радикально выполненное оперативное вмешательство при остеоид-остеоме приводит к стойкому исчезновению болевого синдрома, быстрому восстановлению функции кисти и улучшению качества жизни ребенка.

Ключевые слова: остеоид-остеома; опухоль; дети; хирургия кисти.

Как цитировать

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BACKGROUND

Tumors and tumor-like conditions of bones account for 3.5%–5.2% of all hand diseases [1]. Osteoid osteoma (OO) is a benign bone-forming skeletal neoplasm according to the World Health Organization's histological classification of bone tumors (2020) [2]. In 1935, Jaffe initially described OO as a small spherical tumor, typically <1.0–1.5 cm in diameter, with a distinctive histological appearance [3]. This benign tumor is more prevalent in men, occurring approximately twice as often as in women, and typically affects individuals aged 20–30 years. Further, it is rare in patients aged >40 years [4, 5]. To date, no cases of malignancy associated with this tumor have been reported [6].

OO is commonly found in the long tubular bones of the lower limb, specifically the femur and tibia, in ~50% of patients. Moreover, it may occur in the humerus and forearm bones, affecting 13.3%–31% of patients. In rare cases, OO is detected in the bones of the hand (in 5%–15% of patients), with the carpal bones being the most commonly affected, followed by the metacarpal bones and phalanges of the fingers [3, 4].

In a study by Ghiam, OO was found in the carpal bones of 26 patients, most commonly in the navicular bone and less commonly in the hook and cephalic bones [7]. In another study, Marcuzzi has described 18 cases of wrist and hand OO, with 2 cases in the navicular bone, 2 in the semilunate bone, and 1 in the cephalic bone [8]. Furthermore, Ambrosia has reported 19 cases, including 2 in the cephalic bone, 1 in the hook bone, and 1 in the triangular bone [9].

Jafari has presented a 20-year follow up of the treatment outcomes of 25 patients with hand and wrist OO. The mean age of the patients was 25.2 ± 7.6 years (range: 16–46 years). The most frequent locations were the proximal phalanges (10 cases) and carpal bones (2 in the navicular, 2 in the cephalic, and 1 in the trapezium) [5].

The initial symptom of OO is pain that gradually increases over time. The pain tends to worsen at night and can usually be managed with nonsteroidal anti-inflammatory drugs. If the tumor is located within a joint, it can cause synovitis and contracture, which may mimic primary arthritis [5, 10]. The salicylate test is used in differential diagnosis. After taking salicylate derivatives, pain decreases and completely improves in some cases. This is because of a decrease in the production of prostaglandins and prostacyclins in the affected area [11].

Benign tumors of the hand are often diagnosed late, resulting in inadequate treatment and a high frequency of unsatisfactory results (3%–16%) [1]. To ensure accurate diagnosis, it is crucial to differentiate OO from other benign and malignant tumors of soft tissues and bones, primary chronic forms of osteomyelitis (e.g., Brodie's abscess and Garre's sclerosing osteomyelitis), rheumatoid diseases,

reactions to foreign bodies, bone necrosis, and Kienböck's disease [11].

Neoplasm diagnostics primarily involve radiological examination and computed tomography (CT). However, several authors have reported that on radiographs in standard projections, detecting the tumor "nest" is not always possible [4, 5, 8].

CT is a reasonable option for patients with a clearly marked lesion on radiographs and for those at the diagnostic-search stage. Various modes of CT image reconstruction can provide an objective view of the tumor focus location [4, 5].

MRI is a less-specific diagnostic tool than CT. Diagnostic errors have been reported in up to 35% of cases when MRI is used as the primary test. This is due to the difficulty in clearly identifying the tumor nidus, with reactive changes in surrounding bone and soft tissue structures often being mistaken for inflammation (arthritis, osteomyelitis), malignant neoplasms, and stress fractures [11, 12].

Owing to the natural course of the disease, independent healing of OO is possible. The nidus may mature and remodel for several years, leading to the disappearance of symptoms [13]. Conservative treatment for OO involves the use of nonsteroidal anti-inflammatory drugs; however, this treatment method is not widespread because of its side effects on the gastrointestinal tract during prolonged use. The literature recommends percutaneous drilling of the nidus under CT control followed by thermocoagulation (radiofrequency ablation). Its use has shown good results, with recovery occurring in 90%–98% of cases. However, because of the small size of the tumor and peculiarities of the anatomical area (close location of tendons, neurovascular bundles), performing such treatment on the hand is challenging. The use of this method is limited because the intervention may damage surrounding heat-sensitive anatomical structures [11, 14].

The primary treatment for hand bone OO is surgical intervention, which involves removing the tumor focus either within the unchanged bone tissue or through excision by a single block [5, 11, 12]. Depending on the size of the post-resection defect, bone grafting may not be necessary. Properly executed surgical interventions enable long-term pain management and patient recovery. If the tumor nest is not completely removed, pain may return in the weeks following surgery [5].

During pathomorphological examination, OO appears macroscopically as round or oval shape with a maximum size of 1.5 cm. Under light microscopy, fields of osteoid bone trabeculae with signs of calcification are surrounded by well-vascularized cellular fibrous tissue with single, sometimes grouped, multinucleated giant cells [10].

Although rare, clinical cases of OO have been reported in patients aged <18 years, with the disease localized in the bones of the hand [3, 12, 15–19].

CLINICAL CASE

Patient P, a 17-year-old boy, was admitted to the Clinic of Reconstructive Microsurgery and Surgery of the Hand at the Turner Research Center for Pediatric Traumatology and Orthopedics, Ministry of Health, Russia, complaining of pain in the left hand. The patient reported that the pain has been present for a year and first appeared after a long skiing session. The pain has since increased and mainly occurs at night and after physical activity. Conservative therapy was administered at the patient's place of residence, including wrist joint immobilization. However, no significant pain relief was observed. Pain severity decreased after taking nonsteroidal anti-inflammatory drugs. Radiological examination did not reveal bone trauma. Investigations conducted at the place of residence showed irregular widening of the joint gap, signs of synovitis of the wrist–carpal joint of the first finger of the hand (according to ultrasound), diffuse edema, and aseptic necrosis of the trapezium bone (according to MRI).

Clinical examination of the left hand showed a slight swelling at the base of the first finger and the area of the first carpometacarpal joint. The skin appeared normal in color. Movements in the fingers and wrist joints were preserved, and no signs of carpal instability were noted. However, active abduction of the first finger of the left hand caused pain in the projection of its base, mainly on the palmar surface. Upon palpation of the area, pain increased on the palmar surface of the hand, and instability

of the first carpometacarpal joint were noted. Axial loading on the first finger was moderately painful. No neurological abnormalities were detected, and all hand grips were available. The grip strength of the right hand was 36.8 kg and that of the left hand was 34.0 kg (Fig. 1).

Radiographs of the hands in standard projections did not reveal any structural changes. However, the CT scan of the left hand's trapezium tubercle area showed a delimited rounded area of destruction measuring 0.41×0.47 cm with a sclerotic rim around the periphery and a centrally located bone density inclusion with a diameter of 0.2 cm. The cortical layer of the affected bone's adjacent sections was thinned, and no periosteal reaction was observed. The anatomical relations of the wrist joints, and small joints of the hand were undisturbed. The articular surfaces were clear and smooth, and no bone-traumatic changes were detected (Fig. 2).

MRI showed diffuse trabecular edema of the trapezium bone of the left hand, with an ovoid osteolytic mass of 5 mm in diameter on the palmar surface of the tubercle. PD SPAIR revealed a hyperintense MR signal, whereas T1WI showed an isohypointense signal. The structure of the tubercle of the trapezium bone on the palmar surface revealed an ovoid osteolytic mass 5 mm in diameter, with clear contours, surrounded by a weakly expressed rim of osteosclerosis around the periphery. This pathological formation was mainly characterized by an isohypointense signal on T1WI and PD SPAIR. Additionally, moderately pronounced edema was observed in the paraossal soft



Fig. 1. External appearance of the hands and amplitude of movements in the wrist joint in patient P (17 years old)



Fig. 2. Patient P, 17 years old. X-ray of the hands in anteroposterior projection and computed tomography of the left hand in MPR mode (frontal and sagittal planes) demonstrating the localization of the tumor nest (arrows)



Fig. 3. Patient P, 17 years old. Magnetic resonance imaging of the left hand demonstrating the localization of the tumor nest (arrow)

tissues of the left hand in the corresponding location (Fig. 3).

Based on the clinical and radiological data, OO of the trapezium bone of the left hand was diagnosed. Because of the persistent pain syndrome and the identified pathologic mass in the left hand, surgical intervention was deemed crucial. Surgery was performed under endotracheal anesthesia supplemented with axillary blockade. After placing a hemostatic tourniquet in the middle third of the left shoulder, an arc-shaped incision was made on the palmar surface in the projection of the proximal row of the wrist bones. Then, the short muscles were severed, and the trapezium bone was isolated. The periosteum in the area of the tumor focus location was separated from the underlying cortical layer without any difficulties, and no involvement in the pathological process was observed (Fig. 4a). A radio-opaque contrast marker was placed for the intraoperative determination of the tumor location. An electron-optical converter was used to confirm the presumed location of the tumor. The tumor was then removed as a single block within the surrounding bone tissue, with a fragment size of 1.0×0.7 cm (Fig. 4b). A follow up radiological study confirmed the complete removal of the tumor. The wound was closed using the intradermal suturing technique and immobilized with a soft dressing.

The bone fragment that was removed during surgery was sent for histological examination.

The patient did not experience any night pain, which was a characteristic symptom before the surgery, on postoperative day 1 and thereafter. The patient was discharged from the clinic on postoperative day 4.

Histological examination revealed multifaceted bone trabeculae in the peripheral areas of the sections (Fig. 5a). The central part of the specimen was a clearly delimited focus (nidus) composed of intertwined thin bone beams with a bizarre shape. A pronounced perifocal osteoblastic reaction was observed, along with a few large osteoclastic multinucleated cells (Fig. 5b). The density of bone beams increased toward the periphery of the nidus. Spindle-shaped fibroblast-type cells (with oval-shaped hyperchromic nuclei and light cytoplasm) and rounded cells (multiple accumulation of osteoclasts) were detected in the intertrabecular space

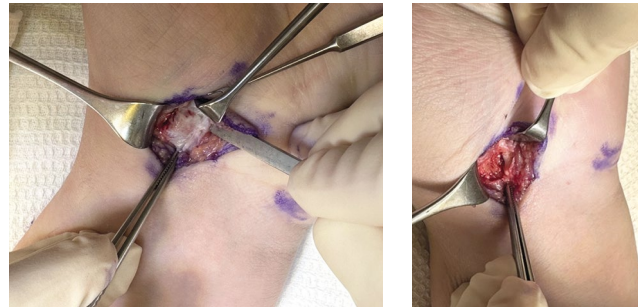


Fig. 4. Patient P, 17 years old. Intraoperative image: *a*, after subcutaneous isolation of the palmar/volar surface of the trapezium bone; *b*, after resection of the tumor "nest" within healthy tissues

(Fig. 5c). The examined material showed predominantly thickened bone trabeculae around the periphery of the nidus, along with pronounced edema and myxomatosis in the adjacent cellular fiber tissue. Moreover, multiple vessels with thickened walls were found in the microcirculatory channel within the cellular fibrous tissue, and in the peripheral sections, sclerosed connective tissue fibers of the periosteum with small and large osteogenic islets were observed (Fig. 5d).

A clinical examination 8 months after surgical intervention showed no signs of disease recurrence. Left wrist joint movements were painless and within the normal range. No pain occurred during physical activity. The grip strength of the right and left hands was 37.0 kg and 35.5 kg, respectively, indicating an increase compared with the initial examination results.

DISCUSSION

Diagnosing OO in the hand can be challenging because of its rarity. One of its characteristic symptoms is nocturnal pain [10]. The discrepancy between severe pain and tumor size may be due to vascularization of the lesion, which increases tension and edema, causing pain through direct stimulation of local nerves around intraosseous vessels. Physical examination typically reveals localized soreness, soft tissue swelling, and decreased range of motion, which can mimic primary arthritis [8]. In this patient population, pain

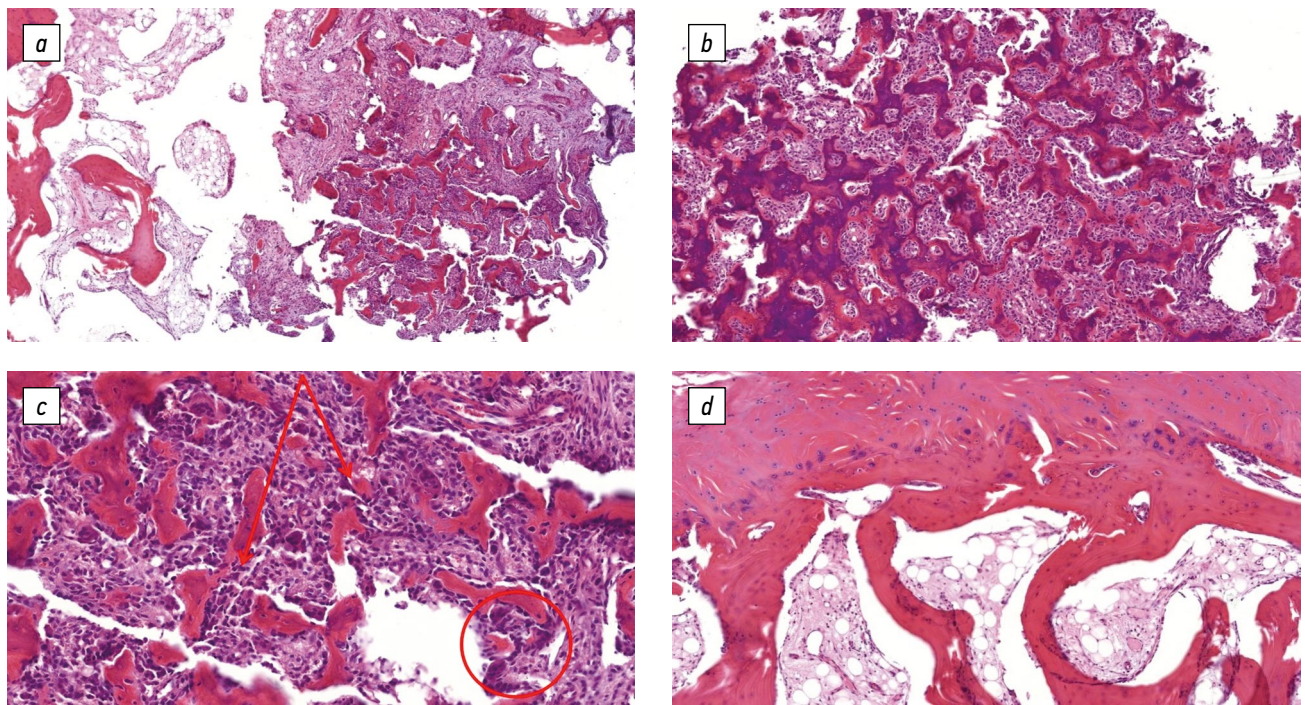


Fig. 5. Patient P, 17 years old. Microphotographs at histological examination: *a*, overview microphotograph showing a section of the tumor nidus bordering adjacent bone tissue, staining with hematoxylin and eosin, $\times 50$; *b*, fragment of the nidus represented by moderately calcified bone beams with cellular fibrous connective tissue in the intertrabecular spaces, staining with hematoxylin and eosin, $\times 100$; *c*, focal osteoclastic reaction with some large multinucleated cells of osteoclastic type, staining with hematoxylin and eosin, $\times 200$; *d*, thickened bone beams bordering with sclerosed connective tissue fibers of the periosteum, staining with hematoxylin and eosin, $\times 100$

sensation decreases with the administration of nonsteroidal anti-inflammatory drugs. Our study patient had a positive salicylate test, which helped to exclude other hand diseases during the examination.

The radiologic picture is typically characterized by the detection of a nidus, a local area of bone destruction up to 1.0 cm in diameter, with an asymmetrically thickened cortical layer [20]. However, radiologic examination did not reveal any bone-traumatic changes, which is consistent with the literature describing initial radiographs of the hand without visible changes [8].

Preoperative CT assessment of bone tissue structural changes aids the surgeon and increases the likelihood of an adequate resection regarding radicality [4, 8].

The tumor did not disrupt the integrity of the cortical layer or periosteum and did not infiltrate the surrounding soft tissues [6], as noted during our surgical intervention.

Several authors have reported positive results in treating patients with OO. After surgical treatment, symptoms regress and the tumor does not recur in remote observations [10, 11]. If the tumor is not completely removed, continued growth may clinically manifest as pain syndrome during the next 7 months after surgical intervention. In most cases, growth continues after neoplasm scraping. Radical surgical intervention is achieved by resecting the tumor in a single block [15].

In our study, diagnosis was confirmed through histological examination, which revealed the characteristic microscopic structure of OO.

CONCLUSIONS

This clinical case demonstrates the importance of comprehensive examination in children with long-term hand pain syndrome. A differential diagnosis of inflammatory processes and excluding other neoplasms is crucial. Surgical treatment can lead to one-stage restoration of hand function and improve the patient's quality of life by providing pain relief.

ADDITIONAL INFORMATION

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Ethical review. Informed consent of the patient for the publication of personal data was obtained. The study was approved by the local ethical committee (protocol no. 23-2; May 29, 2023).

Authors' contribution. *N.V. Avdeychik*, literature analysis, writing the text of the article; *S.I. Golyana*, research concept, editing the text of the article; *D.Yu. Grankin*, search and analysis of literary sources; *A.D. Nilov*, performing histological study, data analysis; *V.V. Chernyavskaya-Haukka*, search of literary sources.

All authors made a significant contribution to the research and preparation of the article and read and approved the final version before publication.

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