Osteoid-osteoma and osteoblastoma with rare localization – presentation of 4 clinical cases

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Abstract— Osteoid-osteoma and osteoblastoma are the two most common representatives of benign bone-forming tumors. Although both tumors are well known to the orthopedic community in Bulgaria, there are several rare localizations that would present difficulties in the diagnostic and therapeutic process. The purpose of our report is to present 4 pediatric cases with rare localization of osteoid-osteoma and osteoblastoma, diagnosed and treated in our clinic in the period between 2020-2022. The presented rare localizations of osteoid-osteoma and osteoblastoma may lead to a delay in making the correct diagnosis. Often, such patients are treated for long periods of time without much effect, which can lead to a deterioration in their quality of life.

Keywords— osteoid-osteoma, osteoblastoma, benign bone tumors

1 Introduction

Osteoid-osteoma and osteoblastoma are the two most common representatives of benign bone-forming tumors. Osteoid-osteoma was first described in 1935 by Jaffe as a benign osteoblastic tumor composed of osteoid and atypical bone. It is a relatively common bone lesion – 3 - 12% of bone tumors according to data from the literature and occurs mainly in childhood and adolescence with a peak in the second decade [1,2]. The most common localization of the tumor is the long tubular bones. The femur and tibia are affected in 75% of cases, spinal localizations reach 10%, and in the bones of the foot the frequency is from 2 to 11% [1,2]. Of these, the talus is the most affected. According to various authors, the patella is affected by less than 1% of all osteoid-osteomas, 0.5%, os ilium – 1%, talus – 1 - 4% (1,2). The most characteristic features of osteoid-osteoma are related to its clinical picture and radiographic characteristics. The presence of a pain, increasing at night and with a rapid effect after the administration of non-steroidal anti-inflammatory drugs, in combination with the presence of an oval leasion with dimensions of 1-1.5 cm and hyperostosis on conventional radiographs, are in many cases sufficient to establish the diagnosis [1,2].

Osteoblastoma is considered by many authors as a "giant" osteoid-osteoma despite differences mainly in the biological behavior of the two nosological units. The tumor accounts for 1-2% of all primary bone tumors [2,3]. The preferred localization is the vertebrae, namely their posterior parts, the facial and cranial bones, followed by the long tubular and epiphysoid bones [2,3]. Metacarpal bones are affected in about 5%, os

ilium – in 2% of cases [2,3]. The tumor grows slowly and is often accompanied by dull pain and a slowly growing swelling in the affected area. Localization in the spine leads to the development of scoliosis [2,3,4]. Radiographically, osteoblastoma appears as an eccentrically located osteolytic lesion with expansive growth and the presence of speckled shadows, most often located in the metaphysis or diaphysis [2,3]. Periosteal reaction in this tumor is rare, and its presence often indicates malignant transformation to osteogenic sarcoma or fibrosarcoma [2,3].

The treatment of both tumors is mainly surgical. Although some cases of osteoid-osteoma can be treated conservatively with NSAIDs, this type of treatment is not recommended in childhood due to the risk of gastrointestinal bleeding and renal dysfunction [5]. Current treatment of osteoid-osteoma is by minimally invasive, CT-assisted radiofrequency ablation. Chahal et al. reported an 89.7% success rate in their series of 87 patients with osteoid-osteoma [6]. Similar results were also reported by Garge et al. in their series of 30 patients [7]. The method is still not widely used in Bulgaria, where open surgery with histological verification is still the standart procedure. Surgical management of osteoblastoma is dictated by tumor grade according to the Enneking Surgical Classification for Musculoskeletal Neoplasms [8]. In most cases, intralesional excision of the tumor, treatment of the bone cavity with mechanical and chemical adjuvants, followed by osteoplasty with a synthetic bone graft is sufficient. In more aggressive lesions, a wide resection with subsequent reconstruction is carried out. The purpose of our report is to present rare localizations of the two described tumors that we diagnosed and treated in our clinic in the period between 2020-2022.

2 Clinical cases

Clinical case № 1:

In the first case, we present a 16-year-old boy with complaints of pain in the left iliac region of about 3 months' duration, worsening at night and quickly disappearing after taking analgesics. A conventional radiograph revealed well-expressed hyperostosis in the left iliac bone and the presence of an oval, well-defined osteolytic leasion (Fig. 1. A). CT was also performed, which confirmed the diagnosis of osteoid-osteoma (Fig. 1 B,C). In this case, open surgical treatment was applied with excochleation of the "nidus" without removal of the entire hyperostosis. The diagnosis was confirmed by histological examination. The postoperative period was smooth, and at the last follow-up, 2 years after the surgical intervention, no recurrence of the tumor was detected.

Clinical Case № 2:

An 8-year-old boy with complaints of pain in the area of the right ankle joint, accompanied by a limitation in the movements, in the last 1 month. Treated conservatively with analgesics without effect. On the radiographs performed, no pathological

changes were detected except for discrete osteosclerosis in the region of the neck of the talus.

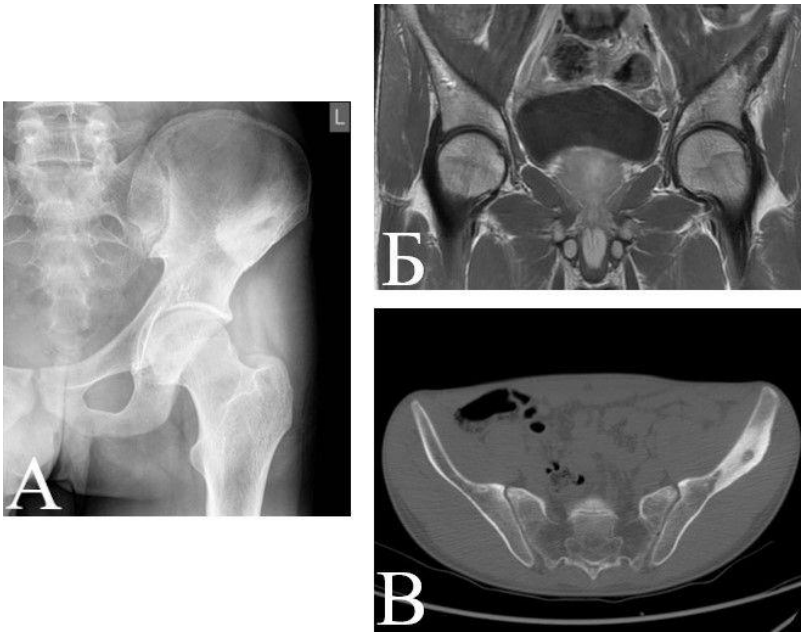

Fig. 1. A – Conventional radiograph showing hyperostosis, in the center of which a well-demarcated osteolytic “nidus” lesion is visible. **B,C** - CT sections that clearly show the exact location of the "nidus".

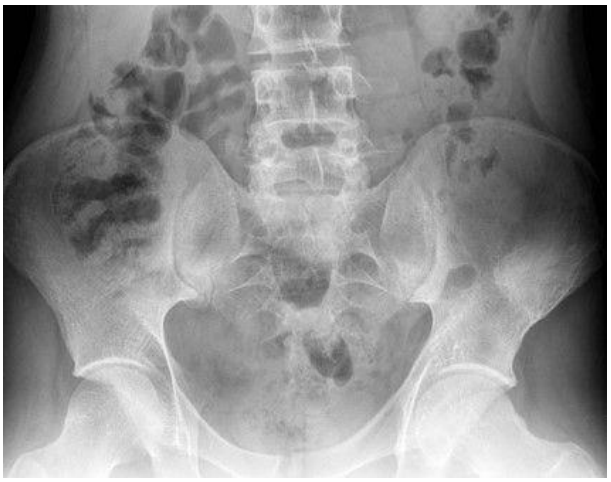

Fig. 2.– Conventional radiograph 2 years after the surgical intervention, without recurrence and with initial resorption of the hyperostosis

This was followed by a CT scan, which visualized a "nidus" in the talus. Similar to the previous case, the treatment consisted of open excochleation of the "nidus", with subsequent histological verification. At the follow-up examination 4 months after the surgical intervention, the child had no pain symptoms and no recurrence of the radiographs performed.

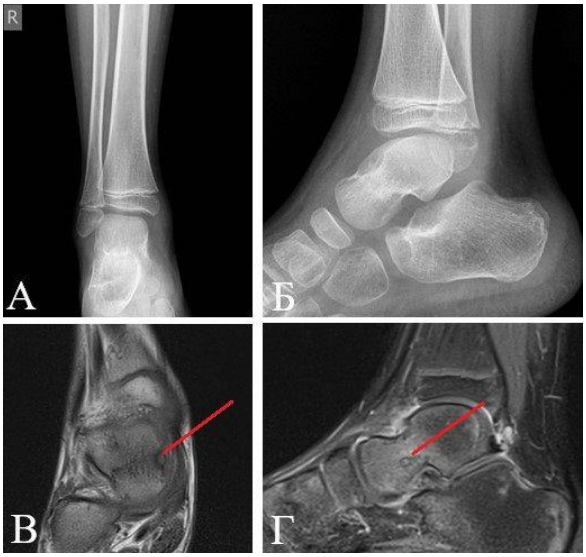

Fig. 3 А,В - Conventional radiograph showing discrete osteosclerosis in the talar neck area, with no "nidus" present. **Б, Г** - CT with clear visualization of the "nidus".

Clinical Case № 3:

A 7-year-old boy with complaints of constant dull pain in the area of the right knee, with a history of about 6 months. No pathological changes were found on the radiographs performed. In contrast, the CT scan showed the presence of a "nidus" in the right patella. The treatment here too was open surgery with removal of the tumor and subsequent histological verification. At the last follow-up, 6 months after the surgical intervention, the patient was without complaints.

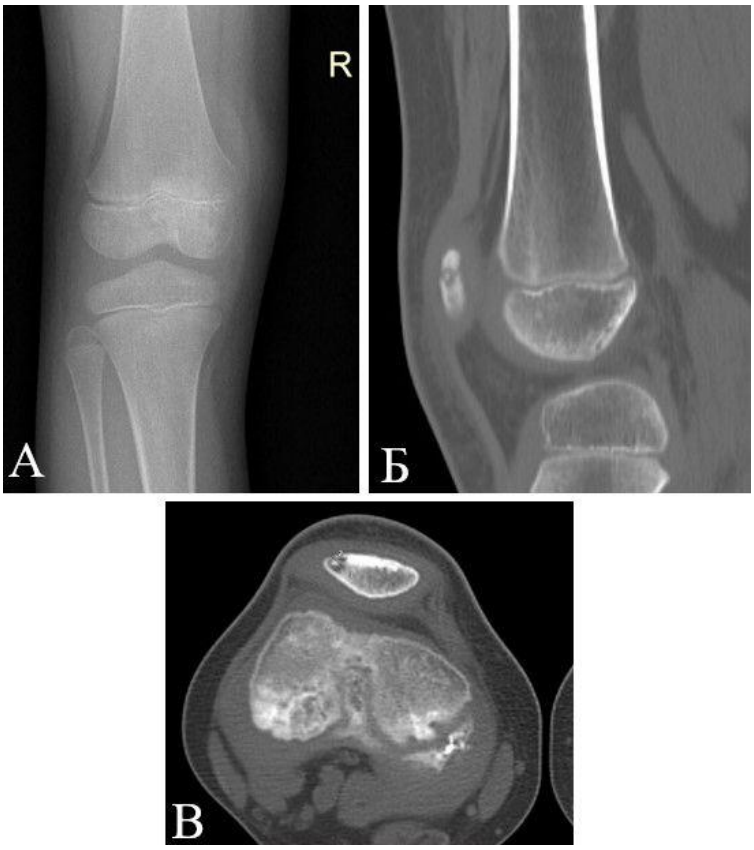

Fig. 4 A – Conventional radiograph without pathological changes. B,C – CT with visualization of the "nidus" in the area of the patella.

Clinical case № 4:

An 11-year-old girl with complaints of pain in the left hip joint, limited movements and a change in gait, in the last 6 months ago. The conventional radiographs performed showed an osteolytic zone in the area of the left acetabulum. This was followed by CT and MRI, which confirmed the presence of a well-demarcated leasion with an osteolytic characteristic and a soft tissue component located mainly in the acetabular fossa. Due to the difficult-to-access location of the lesion and the patient's age, a one-stage surgical treatment was chosen.

Together with colleagues from the Clinic of Pediatric Orthopedics, we performed a Ganz surgical dislocation of the hip, which allowed full visualization of the entire acetabulum. Without injuring the cartilage on the surface of the latter, a careful excision of the formation was performed, which macroscopically was soft, dark reddish and "sandy" pathological tissue. Histologically, the diagnosis of osteoblastoma was confirmed. The rehabilitation regime in this case was standard with off-loading of the limb

for about 6 weeks. At the last follow-up, the patient has no recurrence of the disease, with full range of motion of the hip joint, without pain and other complications.

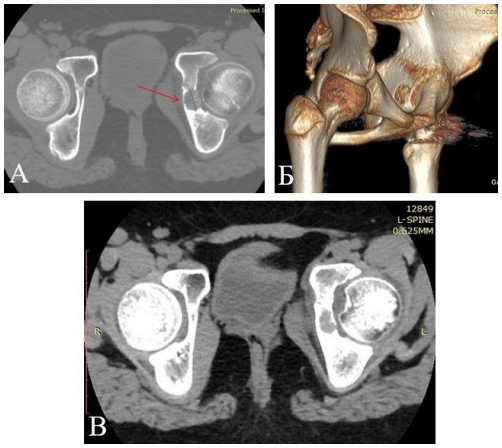

Fig. 5 A, B, C – CT showing the presence of an osteolytic lesion in the region of the left acetabulum.

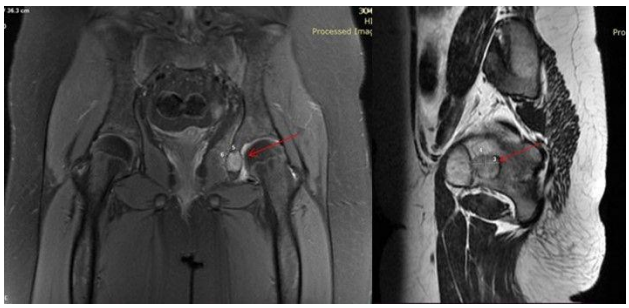

Fig. 6 – MRI showing a T2 hyperintense and T1 hypointense leasion in the region of the left acetabulum.

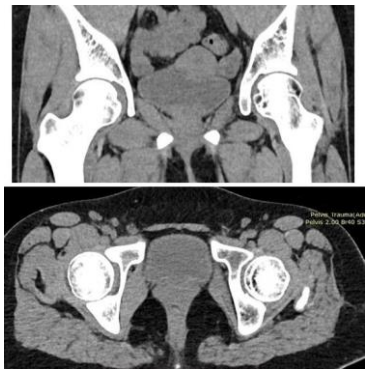

Fig. 7 - Follow-up CT, 2 years after surgery, no evidence of recurrence.

3 Discussion

Although osteoid-osteoma is a well-known benign bone tumor for most specialists in orthopedics and traumatology, the rare localizations presented by us can lead to difficulties in making a correct diagnosis. The rare patellar localization may present as anterior knee pain and is often missed. The combination of a high index of suspicion, appropriate imaging, especially CT, and complete removal of the lesion are conditions for achieving good results. The diagnosis of osteoid-osteoma of the small bones of the hand and foot is difficult. A lesion in the talus may present with signs similar to juvenile monoarthritis, idiopathic arthritis, synovitis, ankle and foot stiffness. Sometimes patients mention a previous trauma, which often delays diagnosis by 2-3, sometimes even 10 years. The differential diagnosis should also be made with osteoblastoma, which is distinguished by its expansive growth and the size of the lesion, which is most often between 3-5 cm in diameter [1,2,3,4]. There is also a rare form of osteosarcoma that originates in the cortex of the bone, called intracortical osteosarcoma, which can mimic osteoid-osteoma early in its development [1,2,3]. The difference here is the dynamic clinical picture and rapid development of the lesion.

Unlike osteoid-osteoma, osteoblastoma is significantly rarer. The localization in the acetabulum described by us is one of the rarest for this tumor. The differential diagnosis here includes osteoid-osteoma, fibrous dysplasia, and low-grade osteosarcoma. [2,3] From fibrous dysplasia, osteoblastoma differs by its expansive growth and the presence of osteoblastic and osteoclast elements on histological examination. Differentiation of osteoblastoma from low-grade osteosarcoma is done mainly by histological examination. In many cases, metastases found in the lungs of patients diagnosed with osteoblastoma are from a misdiagnosed low-grade osteosarcoma. This tumor has relatively slow progression and unspecific clinical presentation.

4 Conclusion

The presented rare localizations of osteoid-osteoma and osteoblastoma may lead to a delay in diagnosis. Often, such patients are treated for long periods of time, without any particular effect, which can lead to a deterioration of their quality of life.

References:

1 Picci P., M. Manfrini, N. Fabbri, M. Gambarotti, D. Vanel, Osteoid-osteoma In: Atlas of musculoskeletal tumors and tumorlike lesions: The Rizzoli case archive, Bologna, Springer International Publishing, Switzerland 2014, 75-77;

2 Dorfman and Czerniak's Bone Tumors, Elsevier Health Sciences. 2015;

3 Picci P., M. Manfrini, N. Fabbri, M. Gambarotti, D. Vanel, Osteoblastoma In: Atlas of musculoskeletal tumors and tumorlike lesions: The Rizzoli case archive, Bologna, Springer International Publishing, Switzerland 2014, 81-83;

4 Galgano, M. A., Goulart, C. R., Iwenofu, H., Chin, L. S., Lavelle, W., & Mendel, E. Osteoblastomas of the spine: a comprehensive review, Neurosurgical Focus FOC, 41(2), E4. Retrieved Jun 26, 2023;

5 Hisaki Aiba, Katsuhiro Hayashi, Hiroyuki Inatani et al., Conservative Treatment for Patients with Osteoid Osteoma: A Case Series, Anticancer Research Jul 2014, 34 (7) 3721-3725;

6 Chahal A, Rajalakshmi P, Khan SA, Rastogi S, Srivastava DN, Gamanagatti S. CT-guided percutaneous radiofrequency ablation of osteoid osteoma: Our experience in 87 patients. Indian J Radiol Imaging. 2017 Apr-Jun;27(2):207-215;

7 Garge S, Keshava SN, Moses V, Chiramel GK, Ahmed M, Mammen S, Madhuri V. Radiofrequency ablation of osteoid osteoma in common and technically challenging locations in pediatric population. Indian J Radiol Imaging. 2017 Jan-Mar;27(1):88-91;

8 Jawad MU, Scully SP. In brief: classifications in brief: enneking classification: benign and malignant tumors of the musculoskeletal system. Clin Orthop Relat Res. 2010 Jul;468(7):2000-2

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