OSTEOID OSTEOMA IN A YOUNG ATHLETE – A CASE REPORT FROM THE PERSPECTIVE OF A PRIMARY HEALTH CARE PEDIATRICIAN

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Summary

Introduction. Osteoid osteoma is a benign osteoblastic tumor, usually 1.5 - 2 cm in diameter, characterized by a well demarcated central area (osteoid nidus) surrounded by a sclerotic bone. It accounts for 5% of all primary bone tumors and most often affects the femur and tibia. The tumor is mostly seen in the second and third decades of life, more often in males, and the typical symptom is pain that worsens at night and responds well to analgesics. Case Report. We present a case of a young athlete, 11 years of age, who presented with characteristic symptoms and localization, as well as a typical radiographic finding. The timely action of a primary health care pediatrician and radiography reduced the time to definitive diagnosis and prompt surgical treatment was provided. The boy gradually started resuming his sports activities six months after the beginning of symptoms and 2.5 months after the surgery. Conclusion. In differential diagnosis of chronic leg pain in patients under the age of 18 years, primary health care pediatricians rely on medical history and physical examination, whereas after that plain radiography of the painful area should be considered. If the nidus is clearly seen in the X-ray, the possibility of diagnosing an osteoid osteoma increases, and timely and adequate treatment are provided. Key words: Bone Neoplasms; Osteoma, Osteoid; Pain; Child; Radiography; Signs and Symptoms; Primary Health Care; Treatment Outcome

Sažetak

Uvod. Osteoid osteoma je benign osteoblastni tumor, obično veličine 1.5 – 2 cm, kogu karakteriše dobro ograničena centralna zona (nidus), oko koje se nalazi zona sklerotične kosti. Čini 5% svih primarnih tumora kosti, a najčešće zahvata femur i tibia. Karakteristično se javlja u drugoj i trećoj dekadi života, češće kod muškaraca, a tipičan simptom je bol koji se pojačava noću i koji dobro reaguje na analgetike. Prikaz slučaja. Prikazujemo slučaj mladog sportiste uzrasta 11 godina kod koga se bolest javila karakterističnim simptomima i lokalizacijom, uz dobru radiografsku vidljivost tumora te je pedijatrijski pregled i pravovremeno radiografsko snimanje značajno skratilo vreme do postavljanja konačne dijagnoze bolesti i obezbedeno je promptno hirurško lečenje. Dečak je 6 meseci od pojave prvih simptoma, a 2,5 meseca nakon operacije uključen postepeno u sportske aktivnosti. Zaključak. U slučaju bola u nozi kod pacijenata starih do 18 godina u primarnoj zdravstvenoj zaštiti, pedijatar se u diferencijalno-diagnoznoj razradi mora osloniti na anamnezu i fizikalni pregled, te doneti odluku o radiologiji bolnog područja. Tako se povećava mogućnost postavljanja dijagnoze osteoid osteoma ukoliko je nidus jasno radiografski izražen. Osobitost ovog bolesti je to da je dovoljno jako radiografski izražen, te se obezbeduje pravovremeno i adekvatno lečenje. Ključne reči: neoplazme kosti; osteoid osteoma; bol; dete; radiografijska vizija; primarna zdravstvena zaštita; ishod lečenja

This tumor does not have the potential for distant metastases but can lead to functional and esthetic deficits [2, 3]. About 50% of lesions occur in the proximal meta-diaphysis of the long bones, most commonly femur or tibia [4, 5]. OO accounts for 10% of all symptomatic benign bone tumors and 5% of all bone tumors. It mostly occurs between 10 and 30 years of age and affects males 2 to 4 times more often than females [2, 3]. The main symptom is pain that worsens at night and is relieved by non-steroidal anti-inflammatory drugs (NSAIDs). The exact mechanism of pain is unknown but increased production of prostaglandins and increased nerve fiber density are found in the nidus [6]. In particular, prostaglandin E2 (PGE 2) is increased up to 1.000 times in comparison with the normal bone and is presumed to be the cause of severe pain seen in pa-

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tients with lesions [7]. Strong expression of cyclooxygenase-1 (COX-1) and cyclooxygenase-2 (COX-2) in the osteoblasts within OO suggests an important role of COX in PGE production, thus explaining good response to NSAIDs [8].

Case Report

We report a case of an 11-year-old boy who visited the Pediatric Outpatient Clinic, part of the Primary Health Care Center complaining about pain in the right thigh in the past 3 months. He started complaining about the pain after a basketball training which he practiced regularly for 3 years, 3 times a week for 1.5 hours. His medical history showed that he had an annual sports physical exam by a pediatrician a month before the first symptoms occurred, without abnormalities at physical examination, electrocardiogram (ECG) and routine laboratory at that time, normal height of 150 cm and weight of 39 kg, and body mass index (BMI) of 17.33 kg/m². Initially, the pain was non-specific, occurred occasionally and lasted for a short time, but sometimes woke him up at night. During the day he felt pain only if he received an accidental kick in the right foot, even if it was of low intensity (unintentionally, passing by). One month after the pain started, he stopped training, and even walking became difficult. His appetite decreased. The NSAIDs relieved the pain, but for a short time, especially at night. Two weeks before seeking medical attention, the pain worsened at night and sometimes he had to take NSAIDs twice a night. He denied any history of trauma or fever. At the first examination by his pediatrician, he was cautious when walking, pale, and felt severe pain on right thigh palpation (pain was evaluated as 7/8 on the Wong Baker Faces Pain Rating Scale). There was no leg deformity, no local swelling, and the skin above the painful area was normal. The X-ray of the right femur in two directions was performed and showed a homogeneous shadow of 80 x 20 mm in the proximal and medial part of the right femur body. In the upper third part of the described shadow an illumination of 10 mm x 3 mm was seen (Figure 1). OO was suspected and the patient was referred to an orthopedist.

The orthopedic physical exam revealed a muscle atrophy of the right thigh and a difference between the right and left thigh muscle volume of 1.5 cm. Hospitalization was indicated by the orthopedist. Computed tomography (CT) of the right thigh confirmed a nidus and open surgical resection under general anesthesia was performed 2 weeks after the pediatrician’s examination. The patient recovered rapidly and was discharged after 7 days of hospitalization. Weight bearing and physical activities were restricted. The patient was able to walk with crutches with the affected foot touching the ground. Five days after discharge, the pain was completely relieved and he stopped taking NSAIDs. Exercises for muscle strengthening progressively improved the range of motion through eight weeks and he was gradually involved in sports activities afterwards. The histopathological analysis confirmed the diagnosis of OO (dg. Neoplasm benignum ossium longorum extrematis inferioris osteoid osteoma) barely 4 months after the onset of pain. After 12 months of follow up, no local recurrence
was observed and the difference between the right and left thigh volume was less than 1 cm. Nine months after surgery and 13 months since the routine pediatric visit, the patient had normal height and weight (155 cm, 41 kg, BMI 17.07 kg/m²).

Discussion

Osteoid osteoma is a benign tumor that commonly affects male adolescents and young adults. The proximal femur and tibia are the commonest locations of OO. The typical clinical presentation is nocturnal pain that responds well to NSAIDs. However, the pain is associated with various disorders which is one of the reasons why OO may be misdiagnosed and correct diagnosis delayed (median 16 months, range 8 – 36 months) [9, 10]. In children, OO may be mimicking different diseases such as infantile cortical hyperostosis, osteomyelitis, Perthes disease, leg length discrepancy, healing stress fractures, tuberculosis, neuromuscular conditions, as well as malignant tumors (osteosarcoma, Ewing sarcoma) [11]. The diagnosis of OO can be a challenging process especially in young athletes. The etiologic factors may be the overuse injury and muscle strain. Some reports present similar clinical cases in athletes with femoral neck stress fractures and intra-articular OO of the femoral neck that may further delay the correct diagnosis [12]. Other unusual localizations, such as metacarpal, hallux, talus or intra-articular are also associated with delayed diagnosis or misdiagnosis of OO [13–16]. Primary health care pediatricians always have a dilemma whether to perform an X-ray of the painful area or not. Radiography is sufficient for establishing the diagnosis of OO if the nidus is clearly expressed, as in this case, so the initial diagnosis was confirmed barely 4 months after the onset of pain. However, there are vague, more calcified cases where nidus overlaps with the surrounding bone sclerosis on the X-rays. Even in case of clear radiographic findings, CT is a gold standard in the preoperative diagnosis of OO [17]. The therapeutic approach depends on the localization, as well as on the severity of symptoms or a potential deformity. NSAIDs may sometimes lead to withdrawal of symptoms and resolution of the tumor [18]. Unsuccessful treatment with NSAIDs or intolerance to analgesics leads to surgical treatment. Open surgical resection is gold standard, but nowadays, minimally invasive surgical methods, such as percutaneous radiofrequency ablation and percutaneous excision, are also becoming important modalities [19, 20]. Marić et al. showed that modified percutaneous techniques provide fast recovery after the procedure and significantly decrease length of hospital stay compared with traditional surgical methods (2.43 ± 0.53 versus 10 ± 7.79 days) [21, 22]. However, even after complete nidus removal, anatomical changes and sequelae may progress and become symptomatic, so long-term follow-up is therefore essential [23]. Histopathological analysis sets the final diagnosis, and as in our case, confirming the clinical and imaging findings. Removal of nidus leads to resolution of pain, so except during the short post-operative period, analgesia is no longer needed, which was also the case in our patient [24].

Conclusion

In differential diagnosis of chronic leg pain in patients under the age of 18 years, primary health care pediatricians rely on medical history and physical examination, whereas after that plain radiography of the painful area should be considered. If the nidus is clearly expressed, the possibility of diagnosing osteoid osteoma timely and providing adequate treatment increases. In the presented case, the correct diagnosis was confirmed barely 4 months after the onset of pain.

References