

Case Report

Case Report: Intraarticular osteoid osteoma of the hip; a challenging diagnosis

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Abstract

Osteoid osteoma (OO) is a relatively common benign skeletal neoplasm of unknown etiology that is composed of osteoid and woven bone. Although, the clinical, radiological and scintigraphic features of OO have been well described, these features may be misleading or altered in the cases of intra-articular involvement. The acetabulum is a relatively uncommon location for OO with a few number of cases reported up to date. We reported a case of a 28-year-old man with intra-articular OO of the hip, located beneath the acetabular rim. Computed tomography, magnetic resonance imaging and bone scans were all nonspecific and confusing. We could show the nidus with the help of PET/CT. The final diagnosis and surgical excision can be made 6 months after the first on set of complaints. Intraarticular OO can mimic lots of pathologic entities related to the affected joint, and presents a diagnostic challenge and cause a delay in the diagnosis. The delays in the diagnosis and treatment can be avoided with a high index of suspicion. Furthermore, PET/CT is a useful imaging modality to demonstrate the tumor especially in the intraarticular location even in the absence of a characteristic nidus. Treatment should be individualized according to the surgeon's preference and the characteristics of the case.

Key words: Osteoid osteoma, hip, acetabulum, positron emission tomography (PET/CT)

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Introduction

Osteoid osteoma (OO) is a relatively common benign skeletal neoplasm of unknown etiology that is composed of osteoid and woven bone. Theoretically, OO can involve any bone, however there is a site predilection for appendicular skeleton, and more than 50% of the cases occur in the tibia and femur [1]. OO is categorized into three types based on the radiographic localization of the nidus; cortical OO, cancellous OO (also referred to as medullary) and subperiosteal OO. Cancellous and subperiosteal OOs typically arise in an intra-articular or juxta-articular location [2]. Although, the clinical, radiological and scintigraphic features of classical cortical OO have been well described, these features may be altered in case of intra-articular involvement. Non-specific clinical manifestations, absence of characteristic radiological and scintigraphic findings may cause a delay in diagnosis and treatment.

Intra-articular OO of the hip located in subchondral bone of the acetabulum is a relatively uncommon lesion with a few number of cases reported up to date [3, 4]. Herein, we described a patient with intra-articular OO of the hip, located beneath the acetabular rim. The purpose of this case report is to illustrate the difficulties in the diagnosis of intra-articular OO and underline the role of imaging modalities in the patients with chronic hip pain and limited joint movements. Furthermore we discussed the treatment options with the review of the current literature.

Case

A 28-year-old man was referred to our clinic with a history of left hip pain for duration of three months. One month ago, he had admitted to another medical center with the same complaints. Conventional pelvis radiograph was taken, however it revealed no abnormal findings (**Figure 1**). The physician thought about the avascular necrosis of the femoral head and ordered hip joint MRI as next step diagnostic work-up. MRI showed nonspecific bone marrow edema of acetabulum and femoral head (**Figure 2**). Thereafter, pelvis CT and bone scintigraphy were taken to get further information about the lesion. Pelvis CT revealed no abnormal findings (**Figure 3**). The increased uptake in left acetabulum and femoral head in bone scintigraphy was interpreted to be compatible with infection (**Fig 4**). Consequently, the patient was treated with antibiotics. After 15 days of antibiotherapy with Piperacillin/Tazobactam, no improvement observed in patient's clinical picture. They decided perform

ultrasound guided fine needle aspiration biopsy from the lesion. However, histopathological examination showed nonspecific inflammatory granulation tissue without a definitive diagnosis.

Figure 1. Pelvis antero-posterior x-ray of the patient on initial admission. No abnormal finding was observed

Figure 2. Images on coronal and axial planes with T1 (TR=660, TE= 13) on the proximal row (a & b) and STIR (TR=2900, TE=30, TI=156) on the lower row (c & d). At the acetabular aspect of the hip joint and on the fovea capitis of the femur, bone marrow edema is observed with their high signal intensity on fluid sensitive sequences and low intensity on the corresponding T1-weighted images. Also note neighborhood muscle edema mainly in the obturatorius internus and abductor muscles.



Figure 1.

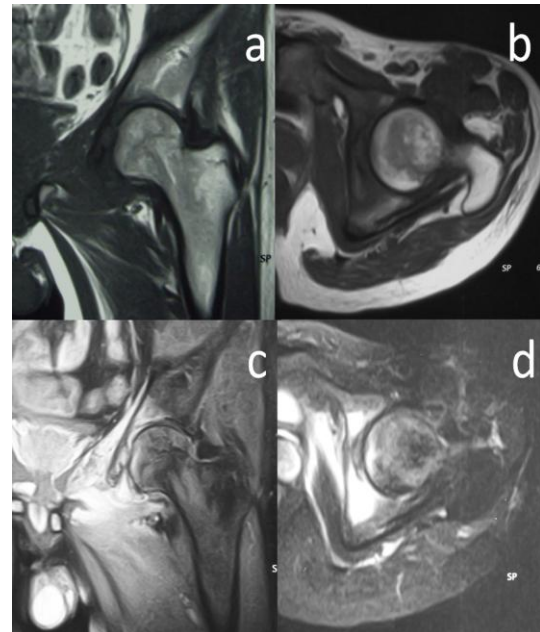
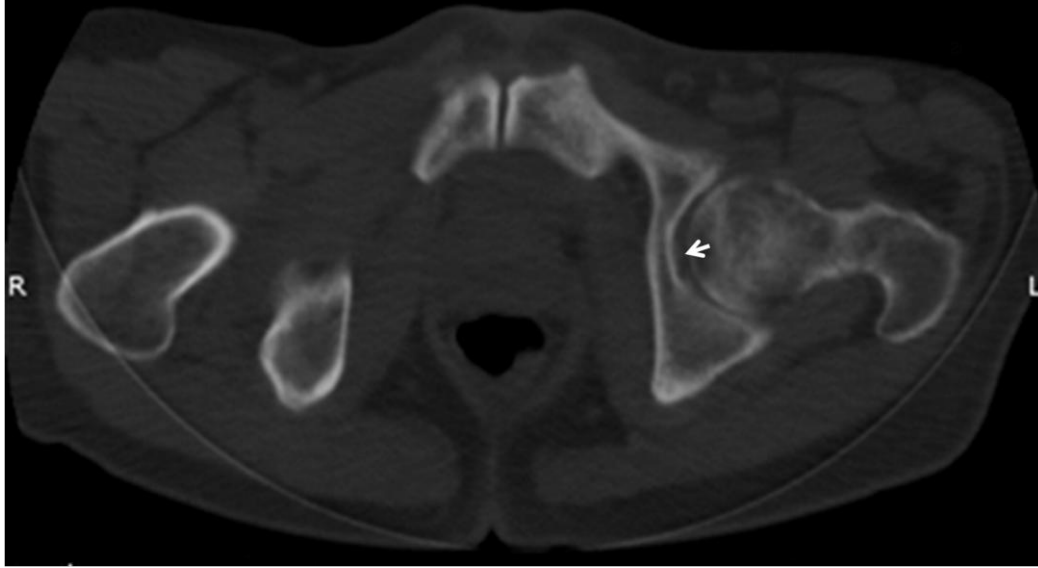


Figure 2.

Figure 3. Axial CT image through the hip joints with 5 mm of slice thickness. An elevation at the center of the acetabular fossa without any visible sclerosis in the bone is seen (arrow).



The patient then referred to our clinic for further evaluation. His initial symptoms were worsened over several weeks, and he reported resting pain at the presentation. Physical examination revealed reduced left hip range of movements in all planes, with end-range pain in all directions. There was 6 cm atrophy of the left thigh compared to the left. Neuro-vascular examination was normal. Routine laboratory studies, including a complete blood count, erythrocyte sedimentation rate (ESR), albumin, alkaline phosphatase, total bilirubin, blood urea nitrogen, calcium, cholesterol, creatinine, glucose, phosphorus, serum glutamic-oxaloacetic transaminase, total protein, and uric acid were all within normal limits. C-reactive protein (CRP) was slightly elevated (10.4mg/L, upper limit 6mg/L). Based on these clinical and imaging findings, other diagnoses including inflammatory arthritis, specific bacterial infections such as tuberculosis and brucellosis were also investigated. PPD, tuberculosis complex PCR, Rose Bengal test, Wright agglutination test, VDRL, HLA-B27, ANA and AMA all yielded negative results. An incisional biopsy was performed from the acetabulum. No pathologic correlation of a specific infection or a neoplastic lesion was established. Electromyography and lumbosacral MRI revealed lumbar nerve root compression and multi-level disc bulging. Therefore, patient received a physical therapy and rehabilitation, and in the meantime femoral and sciatic nerve blocks were performed. However, symptoms did not improve. For the ongoing symptoms, Fluorine-18-2-Fluoro-2-Deoxy-D-Glucose Positron Emission Tomography (FDG-PET)/ CT scan was performed and showed a focal area of

increased uptake in the medial aspect of the left acetabulum, adjacent to the insertion of the ligamentum teres (**Figure 5**).

Open surgical excision of the lesion was performed by anterior Smith-Petersen approach to the hip. A greater trochanter flip osteotomy was made, and the greater trochanter was retracted anteriorly along with the vastus lateralis and the gluteus medius to expose the capsule. A z-shaped capsulotomy was made protecting the lateral retinacular arteries, and the hip joint was subluxated or dislocated anteriorly by flexion-external rotation-adduction. There was a damaged focal cartilaginous lesion over the acetabular cavity. The lesion was completely excised with an adjacent normal bone. The postoperative course was uneventful. The patient was mobilized with crutches in the first day after surgery. Histological examination confirmed the diagnosis of osteoid osteoma. Immediately after the recovery period 1 month after the operation the hip pain disappeared. At the final follow-up 12 months after the surgical excision, the patient was still free of pain with full-range of hip movements, and ambulating unaided. Pelvis CT showed a good remodeling of the curetted lesion (**Figure 6**). Written informed consent was obtained from the patient to present medical data and imaging findings.

Figure 4. Bone scintigraphy of the patient. The increased uptake in left acetabulum and femoral head is seen.

Figure 5. (FDG-PET)/ CT examination of the hip. A focal area of increased uptake in the medial aspect of the left acetabulum, adjacent to the insertion of the ligamentum teres is clearly seen.

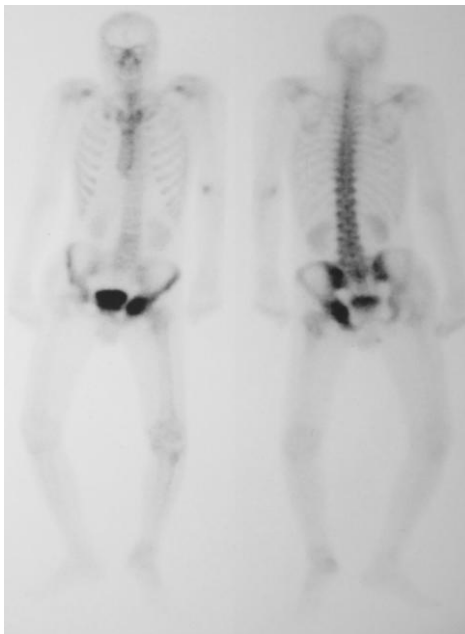


Figure 4.

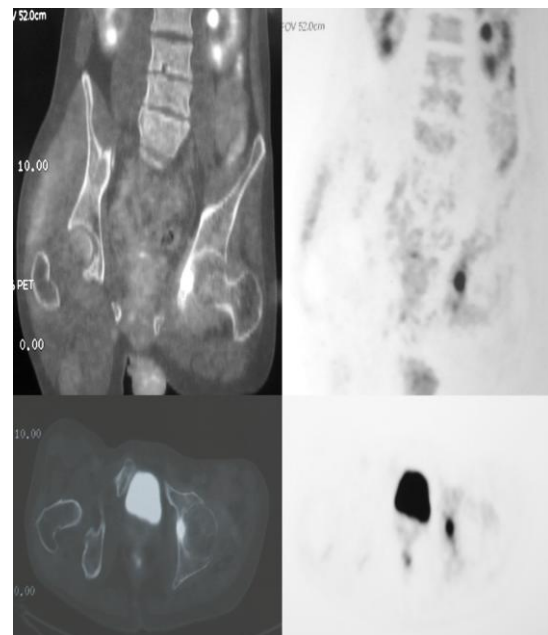
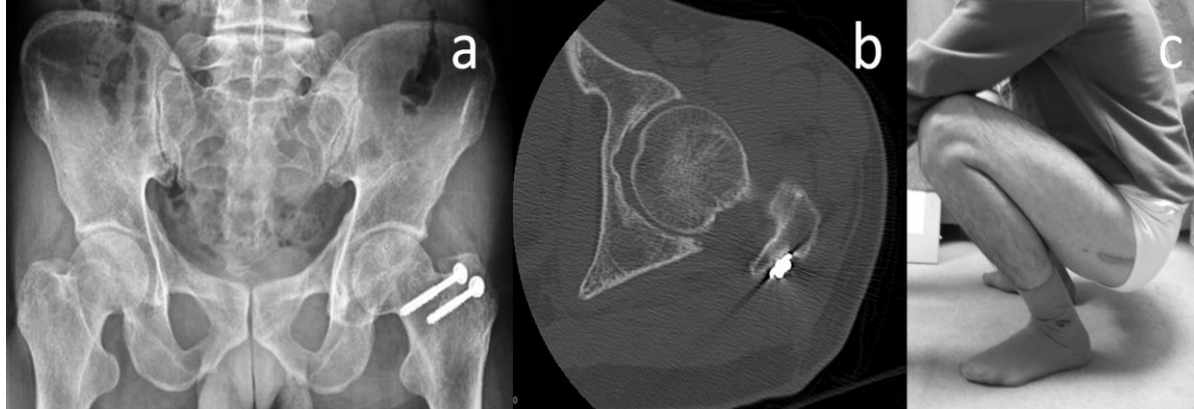


Figure 5.

Figure 6. Final follow-up 12 months after the surgery. (a) Pelvis anteroposterior x-ray of the pelvis, the trochanteric osteotomy united. (b) Pelvis CT shows no recurrence a good consolidation of the curetted lesion. (c) Clinical appearance of the patient.



Discussion

An OO is a solitary, benign bone-forming tumor that is usually less than 15mm in size. It is most commonly seen in the long tubular bones of the lower extremities of patients in the second and third decade. Patients typically present with pain at night and with pain relief by use of NSAID's. The pain usually becomes constant and with increasing severity in time [1]. The high levels of prostaglandins within the nidus and the unmyelinated nerve fibers identified in the fibrous zone around the central nidus or in the nidus itself are believed to be the cause of pain [5]. This is the reason why NSAID's can dramatically relieve the pain. In the vertebrae, OOs usually arise from the posterior elements such as pedicle or spinous process and may cause painful scoliosis or torticollis [2]. On the other hand, the classical nocturnal pain found in around 80% of extra-articular OOs is absent and the pain is less responsive to salicylates when the lesion is intra-articular [1, 6]. Intra-articular OOs are always characterized by signs of synovitis. Some authors suggest that the cause of synovitis and nonspecific arthritis is prostaglandin release from the nidus [7, 8]. The symptoms related to that joint can be confusing and physicians usually think other intra-articular pathologies in the differential diagnosis.

The radiological findings are also atypical and confusing in intra-articular OO of the hip. Intra-articular tumors of the bony pelvis are difficult to diagnose with plain radiographs, not only due to the absent or minimal surrounding osteosclerosis, but also the anatomic

complexity of the bony structure and superimposition [3, 9]. A pelvis radiograph may reveal nonspecific findings of joint space widening, joint effusion and sclerosis, however a characteristic radiolucent nidus may not be present. Similarly in the presented case, a plain radiograph of the pelvis was inconclusive.

Fine-cut CT imaging is the best method of identifying the nidus when the lesion is small or not apparent on plain X-ray particularly in the spine, pelvis and femoral neck or in intra-cortical lesions [2, 10]. However, formation of the nidus may take a longer time in cancellous bone, thus CT may not provide pathognomonic findings. On the other hand, the excellence in soft tissue imaging makes MRI an alternative radiographic method for evaluating joint disorders and demonstrates the tumor nidus of the intra-articular OO. Furthermore, it provides valuable information to rule out the other possible intra-articular pathologies. But the presence of secondary synovitis and marrow edema may lead to erroneous diagnoses [11]. The sclerotic bone surrounding the nidus demonstrates low signal intensity on T1- and T2-weighted images. Marrow edema can be identified on STIR or fat-suppressed T2-weighted fast spin-echo sequences.

Technetium bone scanning has high sensibility but low specificity [10]. It commonly shows a double-density sign with increased uptake centrally that distinguishes the lesion from osteomyelitis or abscess. This sign is characterized by a focal hot spot of the nidus and an area of low peripheral radionuclide accumulation around it, which is related to the sclerotic bone. Unfortunately, in intra-articular lesions, this sign can be absent probably because of reactive hyperemia and associated synovitis [12].

Because of the nonspecific symptoms and the lack of characteristic radiological features of intra-articular OO, there are up to 2.5-3.5 years delays in the diagnosis and treatment in the published cases [1]. In our case, the patient's diagnostic process took almost 6 months after the first on set of complaints. A fine needle biopsy and an incisional biopsy were performed, to reach a definitive diagnosis. However, pathological examination was also inconclusive. We believe that we could not take a proper sample through the lesion. In this delaying and challenging diagnostic pathway, the pathologic entities that can cause similar symptoms and signs should be taken under consideration in differential diagnosis. These include acute, subacute and chronic inflammatory arthritides like septic or tuberculous arthritis, pigmented villonodular synovitis, synovial chondromatosis, gout, rheumatoid arthritis, osteoarthritis, Brodie's abscess and intracortical chondroblastoma [1].

Finally, we referred patient to another medical center for a PET/CT evaluation. 18F-FDG PET is reported to be the most sensitive imaging technique for detection of malignant bone lesions. 18F-Fluoride uptake reflects blood flow and remodeling of bone, which also indicates reactive osteoblastic activity in lytic lesions, even with minimal activity. Because of the limited specificity of this imaging modality, it is not always possible to differentiate benign from malignant lesions, based on the intensity of uptake [13]. Therefore, using of the hybrid PET/CT systems has improved the accuracy of differentiation between benign and metastatic lesions. The occult lesions in anatomically complicated body regions such as the hip joint and pelvis will be overlooked when CT was interpreted alone. The combination of scintigraphic findings with CT in hybrid techniques allowed the accurate identification of these lesions. The major disadvantage of this precious modality is the limited availability of 18F-Fluoride and of PET and PET/CT systems. In our case, we could show the nidus with the help of PET/CT.

The surgical excision of the OO in acetabulum is a difficult procedure. Traditional open surgical approach requires a large incision, wide dissection, hip dislocation and considerable recovery time. Difficulties in accessing the lesion in this localization through open surgery have made the minimally invasive interventions more appealing methods of treatment. Such methods include CT-guided ablation using radiofrequency (RF) and arthroscopic surgery. However, recurrence after RF ablation has been reported to occur in up to ¼ of patients due to incomplete ablation [1]. Furthermore, the procedure is associated with a possible thermal damage to the surrounding neurovascular structures and sometimes it is not possible to obtain a specimen for pathologic examination, thus definitive diagnosis [14].

Arthroscopic excision of OO in the acetabular fossa has been reported [4, 15]. Easy access to lesions in intra-articular locations and cosmetic benefits are the advantages of arthroscopic excision. Limitations of hip arthroscopy can be listed as failure of arthroscopic approach, possibility of nerve injury, and incomplete excision of the lesion. Moreover, it is not a routinely performed technique in many orthopaedic clinics. In our case, we have performed open surgery with trochanteric osteotomy and safe dislocation of hip. Although, technically a challenging procedure which is prone to several complications. We could safely excise the lesion and provide a favorable outcome. One of the reasons why we had chosen the open surgery is the obtaining pathological specimen for the definitive diagnosis.

Conclusions

In conclusion, there are significant differences in the symptoms and imaging features between intra- and extra-articular OO. Intra-articular OO can mimic lots of pathologic entities related to the affected joint. That presents a diagnostic challenge and cause a delay in the diagnosis. As in our case, in the initial phases of the disease, the imaging features may not correlate with the symptoms and a specific finding may not be distinctive. The delays in the diagnosis and treatment may be avoided with a high index of suspicion and patience of the surgeon. The PET/CT is a useful imaging modality to demonstrate an intra-articular OO even in the absence of a characteristic nidus. Treatment should be individualized according to the surgeon's preference and the characteristics of the case. We suggest open surgical techniques in such cases to obtain a pathological specimen, thus reaching definitive diagnosis. This is especially important for documentation and medico-legal purposes.

Conflict of interest: The authors declare that they have no conflict of interest.

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