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Editorial Article

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Osteoid Osteoma of The Lesser Trochanter in Children

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Editorial

The purpose of this editorial is to perform an extensive review of the pediatric literature about the diagnosis and treatment of osteoid osteoma localized in the region of the lesser trochanter, to indicate that it may appear with atypical clinical and/or imaging findings, and to present two illustrative cases, with thigh atrophy and cortical erosion, respectively.

The hip is a common location of osteoid osteoma, with approximately 26% of the total number of cases and two-thirds of the femoral lesions. The tumor is usually intraarticular (intracapsular) and localization in the femoral neck is much more common than in the femoral head or acetabulum [1-3]. It has been previously stated that osteoid osteoma occurring around a joint in close proximity to the capsule, although not in the synovial cavity, may also be considered as intraarticular lesion [4]. These lesions are more properly defined as juxtaarticular (periarticular) [5-8].

Osteoid osteomas located in the region of the lesser trochanter are rare in children and they may appear as intra-, juxta-, or extraarticular lesions. The typical clinical feature of nocturnal pain that is relieved by salicylate (aspirin) or nonsteroidal anti-inflammatory drugs (NSAIDs) is more frequently observed in extraarticular lesions. Diagnosis is especially challenging in patients less than 5 years of age and even more in children that are just beginning to walk [9-11].

Intra/juxtaarticular osteoid osteomas may result in the presenting clinical symptoms of synovitis, and concomitant pathologic changes in the joint surface and synovial tissues (hypertrophic degenerative arthritis) [12]. This nonspecific proliferative synovitis, usually lymphofollicular in nature [13], is a major diagnostic trap because it mimics inflammatory synovitis of the hip, although erythema and warmth are almost always absent [14-17]. Synovitis and joint effusion have been detected in intra/juxtaarticular lesions with an incidence of 73% [18]. However, they have also been reported in 10.6% of the extraarticular lesions, but without presenting a sufficient scientific etiology [19]. In addition, referred pain to the medial aspect of the homolateral knee has been reported to arise commonly from lesions located in the region of the lesser trochanter in children, although a description referring to the anatomic relation of the tumor with the hip joint capsule was lacking [20, 21].

A limping gait, due to restricted mobility of the hip joint and/or pain, as well as neurological signs may occasionally be the presenting complaint of both intra/juxtaarticular and extraarticular hip lesions in children [16, 17, 22, 23]. Patients with hip or femoral lesions may present with thigh atrophy and occasionally with calf wasting [22, 24]. Muscle atrophy usually leads to a wider differential diagnosis [25]. Subsequently, proper diagnosis requires a detailed history and clinical awareness of this phenomenon [21, 22]. Neurological manifestations mimicking spinal root compression have also been reported in children suffering from osteoid osteoma of the hip [21, 26]. Diagnostic testing aims at excluding potential neurological etiologies, causing a delayed diagnosis or misdiagnosis of a neurological disorder [27]. Findings, such as muscle atrophy, weakness, contractures, and diminished stretch reflexes that may be encountered in the process of osteoid osteoma, necessitate a thorough neurological evaluation [28]. In childhood, various diseases may be complicated by different degrees of muscle wasting in the involved extremities, and there are different patterns of muscle wasting of various components of the quadriceps femoris muscle [21, 29-31].

Delayed diagnosis of osteoid osteoma in children may also be explained by the lack of awareness that plain radiographs can be normal in the early stages [32]. Radiographic findings may also be different between intra-, juxta-, and extraarticular osteoid osteomas. The typical plain radiographic feature of a cortically based round or ovoid, radiolucent or partially calcified nidus surrounded by a sclerotic margin with adjacent periosteal reaction is commonly present only in extraarticular lesions. Radiographic follow-up after surgical treatment shows that these changes can be reversible, especially when diagnosis and treatment are made early [33]. In intra/juxtaarticular osteoid osteomas of the hip, the perifocal sclerotic reaction may be minimal or absent, and the periosteal reaction may be subtle due to functional differences of the periosteum of the

femoral neck [4, 34]. However, periosteal reaction may be evident at a remote site, outside the limits of the hip joint capsule, even on both sides of the femoral diaphysis [35, 36]. The appearance of the nidus may also be atypical, presenting as regional osteoporosis of the hip [18, 37, 38]. Cortical erosion overlying the nidus is an extremely rare imaging feature, which has been identified only in phalangeal osteoid osteomas [39, 40].

Computed tomography (CT) is usually confirmatory and remains the investigation of choice for the identification of the nidus. It may also provide a precise anatomy of the area around the nidus, and may help in therapeutic decision making and surgical planning [41]. However, magnetic resonance imaging (MRI) is preferred in pediatric patients due to being radiation-free, and since osteomyelitis and bone tumors are included in the differential diagnosis. MRI may show a variety of findings depending upon the age of the lesion. It may indicate synovitis and joint effusion, bone marrow and soft tissue edema, osteopenia, and muscle atrophy in the affected extremity [42-45]. In patients with hip synovitis, joint effusion, and bone marrow edema, when no obvious tumor can be detected, the diagnosis of osteoid osteoma should be at the top of the differential diagnosis, if the patient has typical pain symptoms [19]. In such cases, care must be taken to avoid erroneous diagnoses and incorrect operative procedures [46-49]. MRI has also been used to assess the impact of osteoid osteomas of the hip on the size and fatty infiltration of the thigh musculature. No significant association was demonstrated with pain duration suggesting that muscle atrophy may rather be related to the locoregional inflammation than subsequent to the disuse of the limb [50].

The 'double-density sign' on bone scan is the classic scintigraphic finding of osteoid osteoma of the appendicular skeleton. However, in intraarticular lesions, this finding is less frequently visible due to less or absent osteosclerosis, and it has been reported to be accurate in only two-thirds of the cases. Scintigraphy may also be used intraoperatively to localize the tumor and to establish complete removal of the nidus [51-57]. An arteriogram may also be useful in intraarticular osteoid osteomas, presenting with scanty bone sclerosis, to indicate the radiographically unrecognized nidus [58].

The clinical differential diagnosis of hip osteoid osteoma ranges between inflammatory and noninflammatory arthritis of the hip, aseptic osteonecrosis of the femoral head, slipped capital femoral epiphysis, radicular syndrome, stress fracture, and regional osteoporosis. The radiographic differentiation includes osteoblastoma, Brodie's and intracortical abscess, chronic osteomyelitis, callus formation due to an avulsion fracture of the lesser trochanter, eosinophilic granuloma (Langerhans cell histiocytosis), bone island, and malignancy [1, 59-69].

Various complications, including growth disturbances as well as bone and joint deformities, have been reported in children suffering from osteoid osteoma of the lower extremities [70-76]. Muscle atrophy and leg length discrepancy have been reported to resolve after treatment [22]. Chronic synovitis due to intra/juxtaarticular osteoid osteoma of the hip may lead to irreversible cartilage destruction [77] and osteoarthritis [78]. The inflammatory and degenerative changes may resolve after surgery, depending on whether the removal of the nidus has been considerably delayed [1].

Epidemiology suggests that osteoid osteoma is self-limiting. A medical approach could be considered if the lesion is clinically well tolerated. Resolution during medical treatment can be associated with a gradual disappearance of the MRI visibility of the nidus and neighboring edema. The purpose of surgery is to eradicate the nidus. Complete surgical excision (en-bloc resection or curettage) has historically been the treatment of choice. However, surgery has disadvantages, including the difficulty of locating the lesion intraoperatively, the potential need for internal fixation and bone grafting, prolonged hospitalization, and the possibility of postoperative complications ranging from an unsatisfactory cosmetic result to a pathologic fracture [79-81]. Deep and no easily accessible osteoid osteomas, like those of the lesser trochanter are associated with a higher risk of complications. An anterior approach is usually preferred over the lateral one because identification of the nidus is easier and there is less risk of targeting error. The use of the posterior and medial adductor approaches has also been reported [82-87]. Accurate intraoperative localization is crucial. Radiography, CT, tetracycline labeling, and bone scintigraphy have all been used for this purpose. The nidus usually appears intraoperatively as a reddish-brown spot [88]. The two most common surgical methods for removing the nidus are en bloc resection and the burr-down technique. The former requires a larger resection of bone than the burr-down technique, and therefore, either bone grafting or internal fixation may be necessary. However, even in cases of successful localization, the surgically created bone defect may lead to an iatrogenic fracture of the proximal femur [89-92].

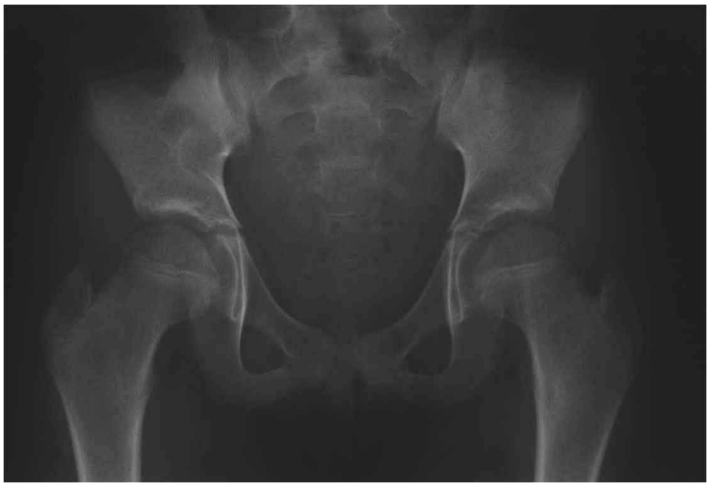
Recurrence is generally considered to result from incomplete resection or destruction of the nidus. The reported amelioration or even complete symptom alteration following removal of the cortex overlying the nidus, leading to an exposed or unroofed nidus [93] has not been confirmed. On the contrary, the surgical dilemma and the fate of patients when no nidus is identified in the resected specimen, indicates that further surgical interventions are required [94].

Minimally invasive techniques have replaced open surgical treatment with favorable results. Their main advantages are lower invasiveness and cost. They include image intensifier or CT-guided percutaneous excision using trephine, and CT- guided percutaneous radiofrequency coagulation [95, 96]. MR-guided focused ultrasound thermal ablation has also been employed with promising results [97, 98]. In addition, arthroscopic excision, radiofrequency thermoablation, laser ablation, and cryoablation have also recently become validated treatment methods, despite the criticism of lacking histological proof for the diagnosis of osteoid osteoma [99-102].

A classification system of osteoid osteomas localized in the region of the lesser trochanter has been attempted by the author on the basis of the anatomical localization of the tumor on the antero-posterior radiographic view. Subsequently, three types of osteoid osteomas occurring in the region of the lesser trochanter may be described. They are all localized medially to the midline of the femoral meta/diaphysis. Osteoid osteomas occurring between the midportion and the medial edge of the intertrochanteric line may be defined as intraarticular lesions. Those localized between the medial border of the intertrochanteric line and the midlevel of the lesser trochanter may be defined as juxtaarticular, while more peripheral lesions, located up to 2 cm below the lesser trochanter, may be defined as extraarticular lesions. The first two types may also be considered as intra- and juxtaarticular osteoid osteomas of the femoral neck, respectively. Although the presence of synovitis and joint effusion is not an absolute prerequisite to define an intra/juxtaarticular lesion, it may be prudent to consider that it is the most secure clinical finding to differentiate between intra/juxtaarticular and extraarticular osteoid osteomas located in the region of the lesser trochanter.

Two illustrative cases with a histologically proven osteoid osteoma of the lesser trochanter are presented. Surgical resection was performed by the author under general anesthesia through an anterior approach. Image intensifier-guided needle localization of the nidus was used in the operating theater in both cases and the tumor was removed en bloc to the margin of reactive bone by placing drill bits around it. The resected piece of bone was x-rayed to secure total removal of the nidus. Persistent lytic areas were evident in the long-term radiographic follow-up in one patient. It may be prudent to consider that there is no need for advanced imaging modalities or treatment of such lytic lesions, as it has already been concluded for pediatric postfracture cystic bone lesions [103] or for cysts smaller than 5 cm following curettage of benign bone tumors around the knee [104].

Patient 1: An 11-year-old girl presented with a 17-month complaint of left hip pain. The pain was intense at night and was controlled by salicylate but became severe, constant, and unaffected by salicylate and other NSAIDs during the last 6 months. Plain radiographs performed about a year ago, were diagnosed with no bone abnormalities (Figure 1A). On admission, a limping gait associated with diminished strength and thigh atrophy of the left leg was diagnosed (Figure 1B). There was local tenderness over the anteromedial surface of the left proximal femur but no local signs of inflammation. Plain radiographs indicated bone sclerosis at the level of the lesser trochanter (Figure 1C). MRI sections showed focal signal abnormality in the anteromedial cortex of the femoral shaft at the level of the lesser trochanter (Figure 1D). There was no evidence of hip joint effusion. The lesion could be defined as an extraarticular lesion since it extended below the midlevel of the lesser trochanter. She reported complete relief of her symptoms postoperatively and muscle atrophy recovered fully within 12 months. Radiographs 5 years postoperatively indicated no regression of the reactive sclerotic bone (Figure 1E). She is symptom free after a 15-year follow-up.



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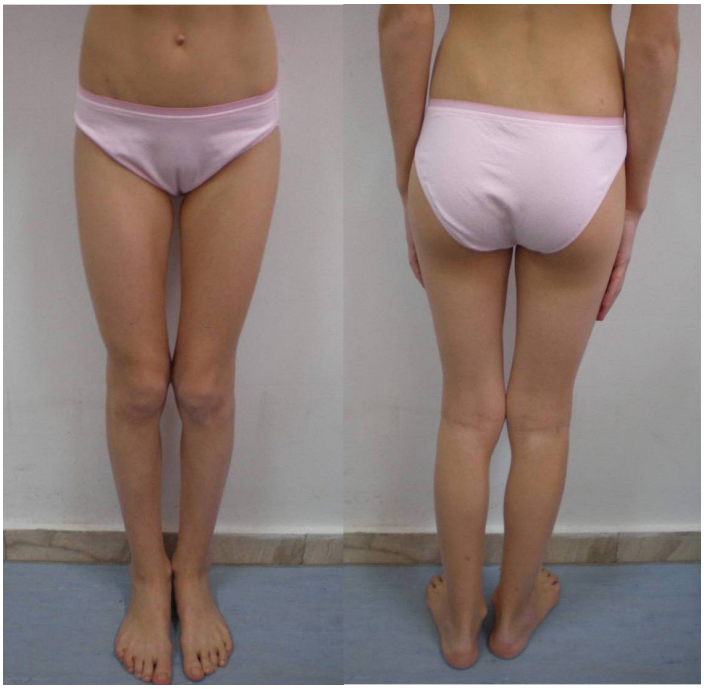

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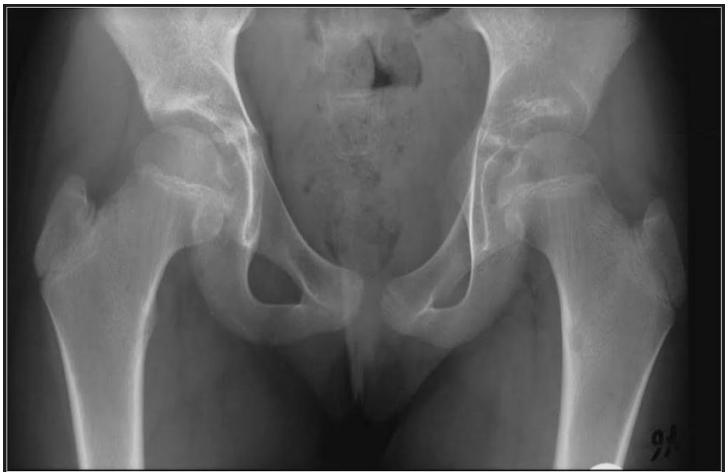

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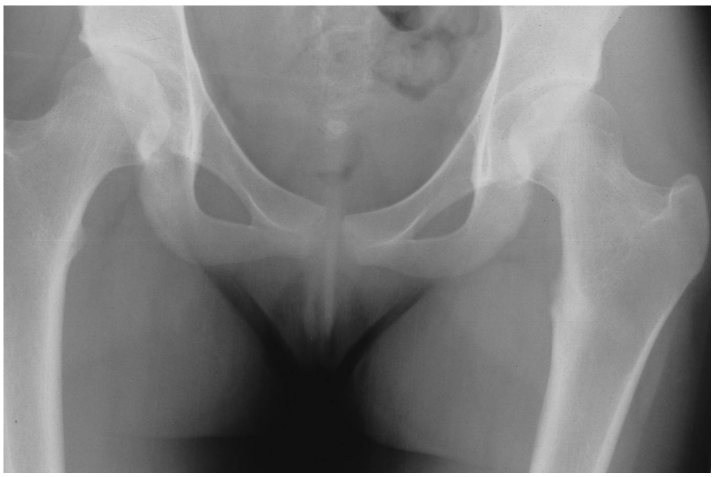

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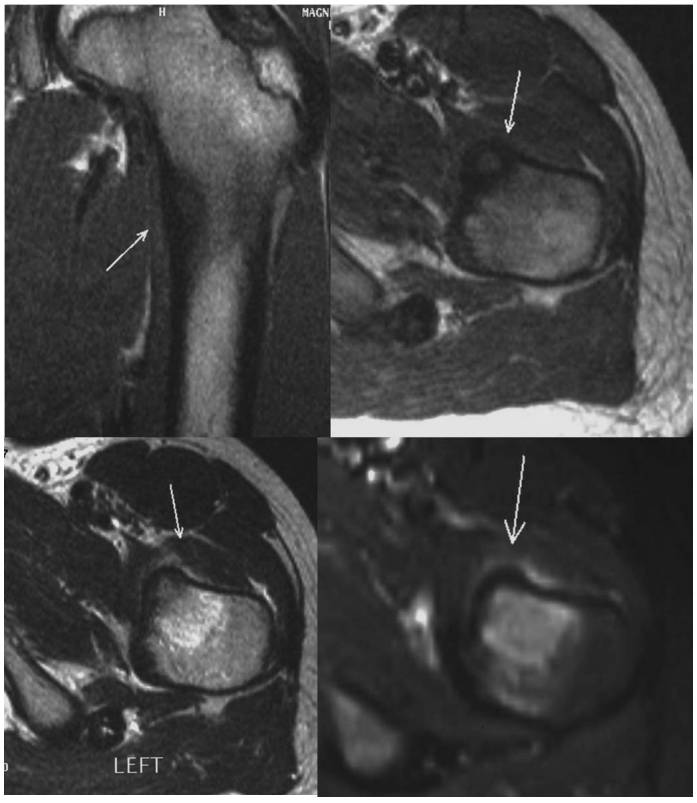
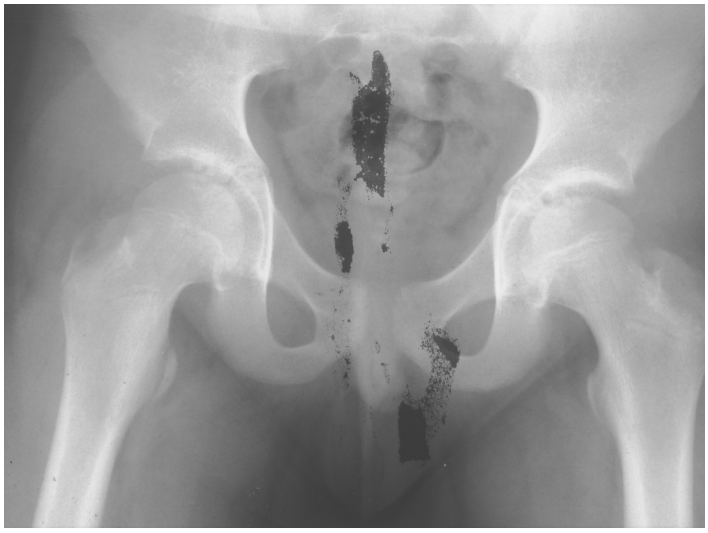


(1D)

Figure 1: An 11-year-old girl with a 17-month history of a painful left hip. Initial radiographs were diagnosed with no abnormal findings (A). On admission, atrophy of the left thigh musculature was evident (B) and radiographs indicated femoral cortical sclerosis at the level of the left lesser trochanter (C). Coronal and axial T1-weighted MR imaging (top views) showed a focus of intermediate signal intensity surrounded by mild reactive sclerosis. Axial T2-weighted (bottom left view) and STIR (bottom right view) MR imaging indicated bone marrow and soft tissue edema (D). Radiograph at 5 years postoperatively indicated persistence of the reactive bone sclerosis (E).

Patient 2: A 13-year-old boy presented with a complaint of 22-month pain of the right hip. The pain was intense at night and was controlled by salicylate but became severe, constant, and unaffected by salicylate and other NSAIDs during the last 10 months. Plain radiographs performed 18 months ago were diagnosed with no bone abnormality (Figure 2A). On admission, a limping gait was evident. There was local tenderness over the anteromedial surface of the right proximal femur. No local signs of inflammation or muscle atrophy were detected. Plain radiographs indicated a lytic bone lesion at the level of the right lesser trochanter with reactive sclerosis and mild periosteal reaction along the medial cortex of the femoral neck (Figure 2B). MRI sections indicated a bone lesion localized in the anterior femoral cortex at the proximal level of the lesser trochanter, 15-16 mm in its biggest dimension. It was surrounded by a fusiform area of increased signal intensity. Bone marrow and deep soft tissue edema were also diagnosed. There was no evidence of hip joint effusion (Figure 2C). CT indicated an 11X10X16 mm lytic intracortical bone lesion with a calcified center. The adjacent cortical bone was thickened and there was pronounced periosteal thickening. Both MRI and CT (Figure 2D) indicated the presence of 3X10 mm cortical erosion overlying the nidus. The lesion could be defined as a juxtaarticular lesion since it was localized between the medial edge of the intertrochanteric line

and the midlevel of the lesser trochanter. Although intraoperative identification of the nidus could be based on the appearance of the cortical erosion, image intensifier-guided needle localization was also used (Figure 2E). He reported complete relief of his symptoms postoperatively. Radiographs 7 years postoperatively indicated no regression of the reactive sclerotic bone and persistent lytic areas (Figure 2F). He is symptom free after a 10-year follow-up.



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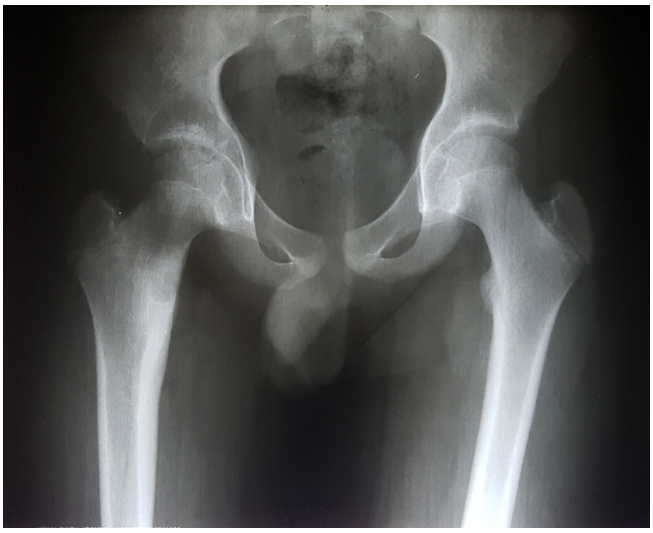

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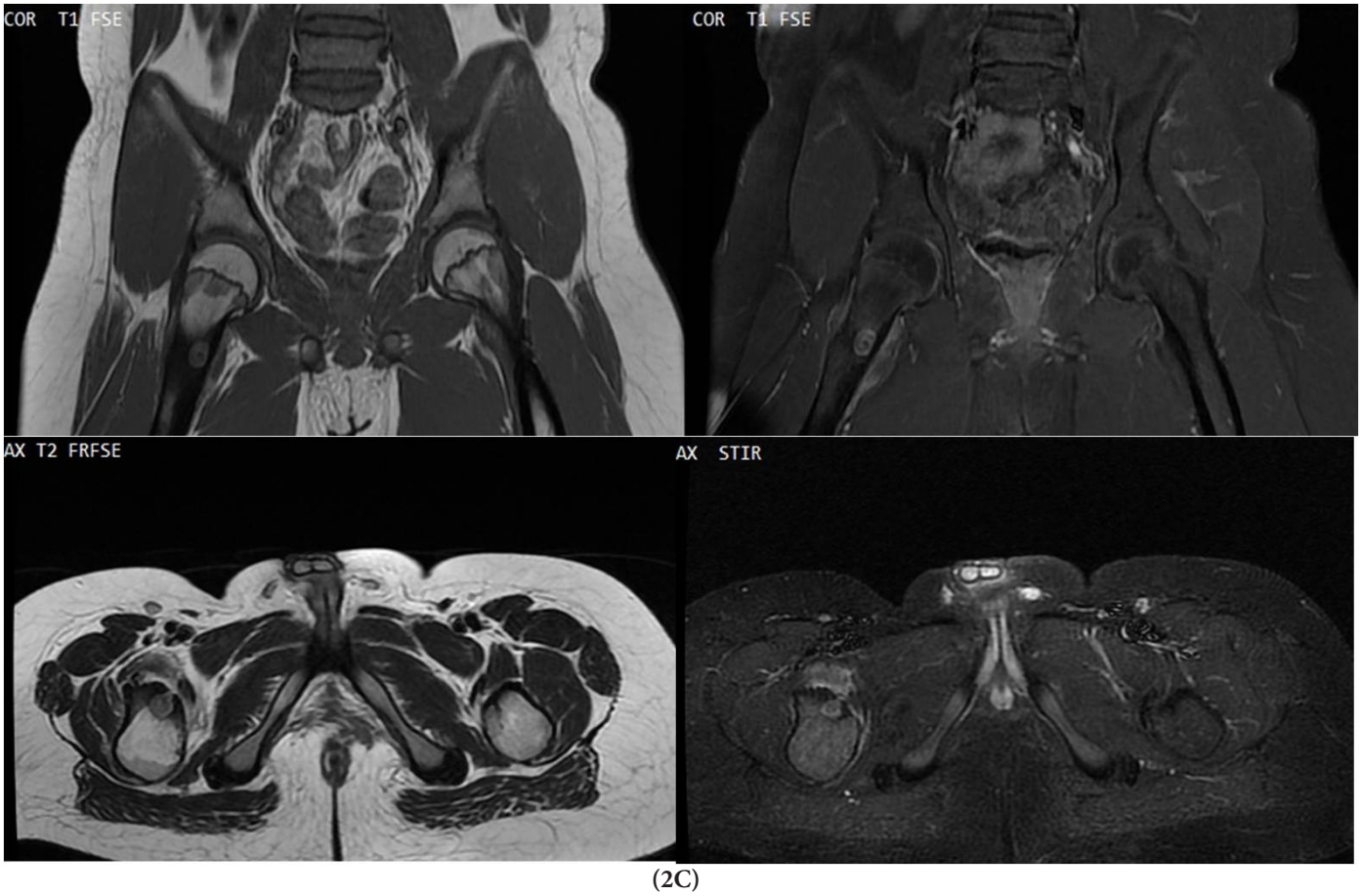

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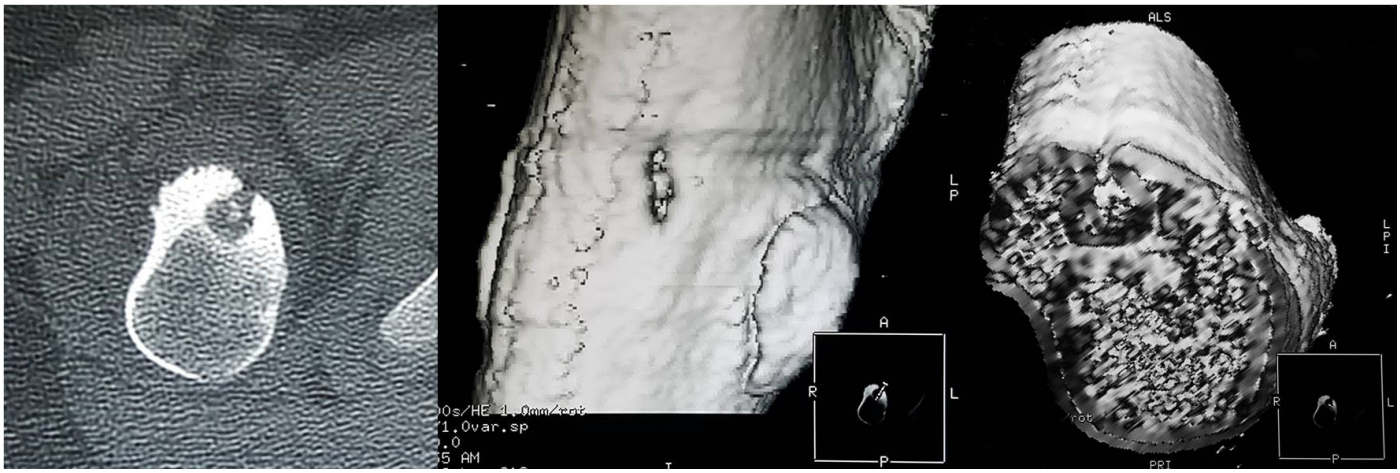

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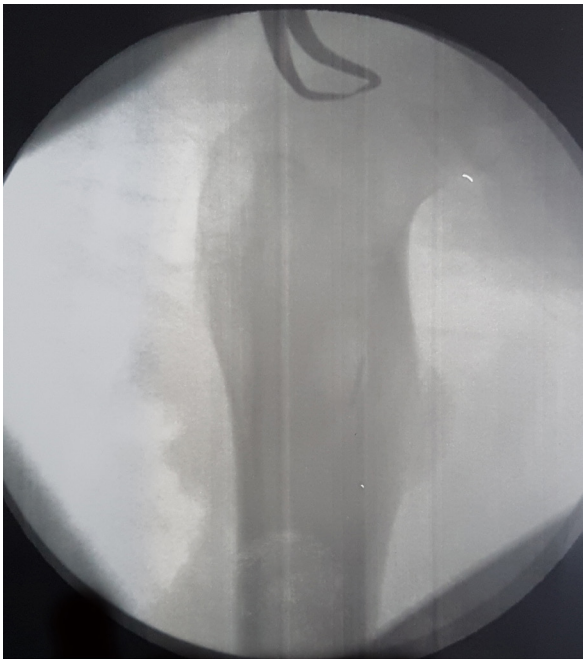

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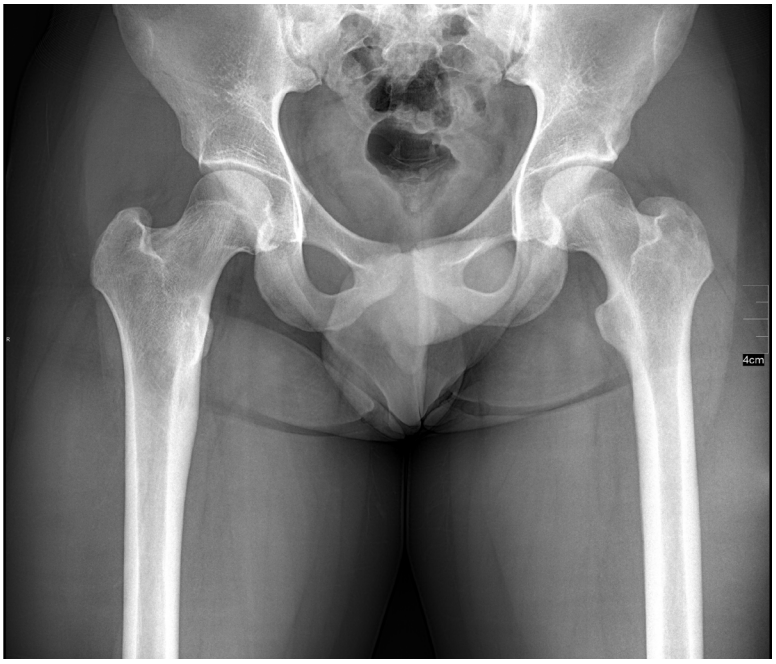

(2F)

Figure 2: A 13-year-old boy with a 22-month history of a painful right hip. Initial radiographs were diagnosed with no abnormal findings (A). On admission, radiographs indicated a lytic bone lesion at the level of the right lesser trochanter with intense reactive bone formation and mild periosteal reaction of the medial femoral neck (B). Coronal T1-weighted FSE (top views), axial T2-weighted FRFSE (bottom left), and axial STIR (bottom right) MR imaging showed a focus of abnormal signal intensity with a calcified centre surrounded by reactive sclerosis, bone marrow and soft tissue edema, and a cortical erosion (C). 2- and 3-dimensional CT indicated a low attenuation area with a calcified center, surrounded by a region of high attenuation reactive sclerosis, and anterior femoral cortical erosion (D). Image intensifier-guided intraoperative needle localization of the nidus was used (E). Radiograph at 7 years postoperatively indicated persistent lytic areas along the medial femoral cortex and reactive bone sclerosis (F).

Conclusion

In children, osteoid osteomas in the region of the lesser trochanter may exhibit either intra/juxtaarticular or extraarticular localization. Both lesions may present with atypical clinical and/or imaging findings. In the two presented children, an extraarticular lesion was associated with thigh atrophy, which fully recovered postoperatively, and a juxtaarticular lesion presented with cortical erosion, an imaging feature that has not been previously reported. En bloc surgical excision of the osteoid osteoma through an anterior approach was performed in both patients with optimal results. Minimal removal of reactive sclerosis with image intensifier-guided needle localization of the nidus as well as confirmation of the totally removed nidus with radiography of the resected piece of bone may still be regarded as a reliable approach. This mode of treatment, when recent minimally invasive techniques are not available, offers histological verification of the diagnosis and may permit no use of internal fixation or bone grafting in children.

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