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Osteoid Osteoma Can Occur at the Pars Interarticularis of the Lumbar Spine, Leading to Misdiagnosis of Lumbar Spondylolysis

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Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
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Conflict of interest: None declared

Patient: Male, 18
Final Diagnosis: Osteoid osteoma
Symptoms: Low back pain
Medication: —
Clinical Procedure: Operation
Specialty: Orthopedics and Traumatology

Objective: Rare disease

Background: Osteoid osteomas are benign bone-forming tumors characterized by local inflammation and pain. They are also characterized by a small osteolytic lesion (nidus). Spondylolysis is a defect of the pars interarticularis, which may lead to stress fractures, and is a common cause of low back pain in adolescence. Osteoid osteoma occurs predominantly in the posterior elements of the spine. Magnetic resonance imaging (MRI) signal abnormality suggesting bone marrow edema is a common finding in osteoid osteoma and early-stage spondylolysis without prominent defect.

Case Report: An 18-year-old male was suffering from low back pain. He was diagnosed with lumbar spondylolysis on initial MRI and computed tomography (CT). Subsequent thin-slice CT demonstrated a nidus at the pars interarticularis, and variously-sliced MRI could detect widespread bone marrow edema. On the diagnosis of an osteoid osteoma, the nidus and surrounding osteosclerosis were resected. The patient's pain disappeared after surgery.

Conclusions: Osteoid osteoma in the pars interarticularis can be difficult to diagnosis, because MRI and CT findings for osteoid osteoma at the pars interarticularis are similar to those of the lumbar spondylolysis. The possibility of osteoid osteoma should be kept in mind when examining adolescents with low back pain.

MeSH Keywords: Spondylolysis • Osteoma, Osteoid • Lumbar Vertebrae • Diagnosis

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Background

Osteoid osteomas are benign bone-forming tumors. The lesions occur in the spine in about 10% of all benign bone tumors, and are predominantly located in the posterior elements [1,2]. The majority of osteoid osteomas occur in the second and third decades of life, with a male preponderance. Osteoid osteomas are characterized by a nidus of mineralized immature bone, associated with a vascularized stroma. The nidus is usually surrounded by osteosclerotic reactive bone [3]. Plain radiographs often fail to depict the nidus as an osteolytic lesion, because the nidus is small. Computed tomography (CT) is useful to show the nidus with varying degrees of perinidal sclerosis [4].

The pain caused by an osteoid osteoma is usually consistent, and is typically worse at night. High expression of cyclooxygenase-2 at the nidus is reported to cause inflammation and pain, and perinidal edema. Nonsteroidal anti-inflammatory drugs (NSAIDs) are known to be effective for relieving the pain of an osteoid osteoma by reducing inflammation [5]. Reflecting the perinidal edema, magnetic resonance imaging (MRI) shows bone marrow edema characterized by high signal intensity on T2-weighted images, which could be the only finding for an osteoid osteoma [6].

Spondylolysis is a defect of pars interarticularis and is found in about 6% of the general population [7]. Spondylolysis is a common cause of back pain in children over the age of 10, especially in young athletes [8,9]. The pathogenesis of spondylolysis is considered to be a stress fracture [10,11]. The early stage of spondylolysis can be divided into very early stage and late early stage [12]. Very early stage defects show faintly on CT, therefore, even using CT, it may be difficult to diagnose a very early stage of spondylolysis [12]. A high signal change in the pedicle on T2-weighted images on MRI could be an indicator of early stage spondylolysis [12]. The abnormal MRI signal in the early stage of spondylolysis is generally indistinguishable from bone marrow edema in the osteoid osteoma in cases in the posterior element of the spine when the nidus fails to be detected.

In the present case report, a teenaged male patient suffered from low back pain with osteoid osteoma in the pars interarticularis of the lumbar spine. The patient was diagnosed as having spondylolysis based upon his clinical symptoms and initial CT and MRI findings. Following detailed image examinations, a diagnosis of osteoid osteoma was made.

Case Report

An 18-year-old male had suffered from low back pain of six-month duration, and visited a nearby orthopedic hospital. A diagnosis of nonspecific low back pain, possibly associated with muscle fatigue was made. Fever and general fatigue were not seen. Medication with NSAIDs effectively reduced the pain. Without the medication, the pain still existed, even

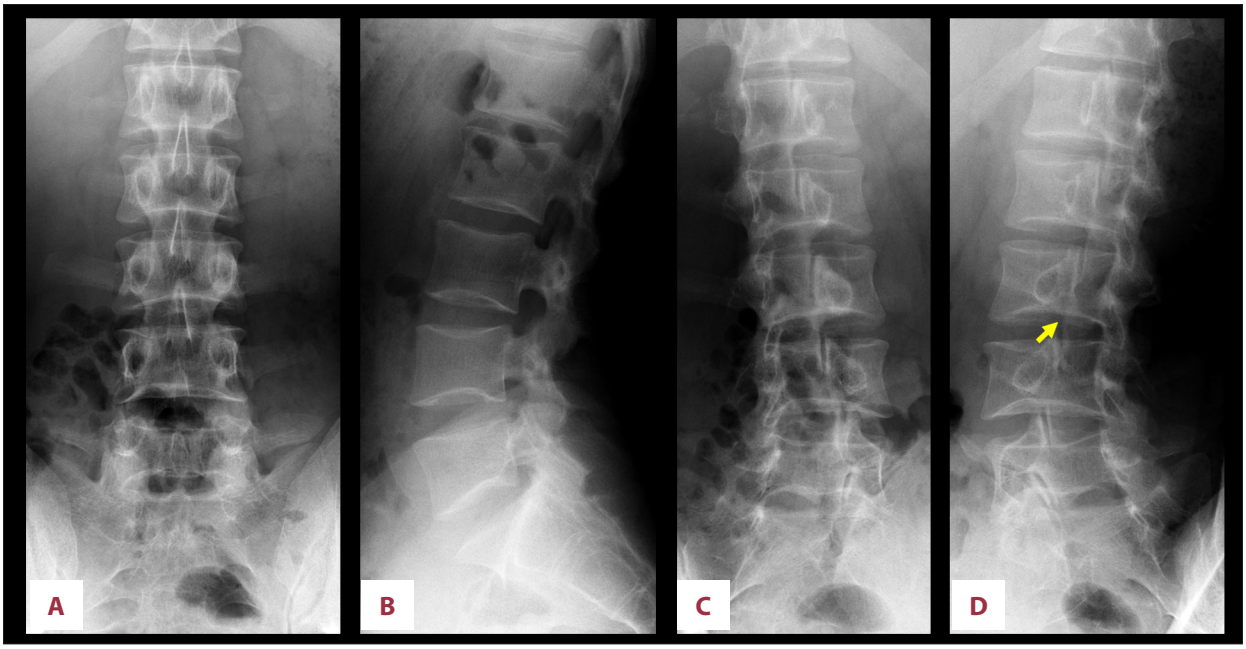

Figure 1. Plain radiographs on the lumber spine look normal on anterior-posterior view (**A**), lateral view (**B**), right anterior-posterior oblique view (**C**), and left anterior-posterior oblique view (**D**). A yellow arrow indicates the place of the lesion detected by the other image analysis (**D**).

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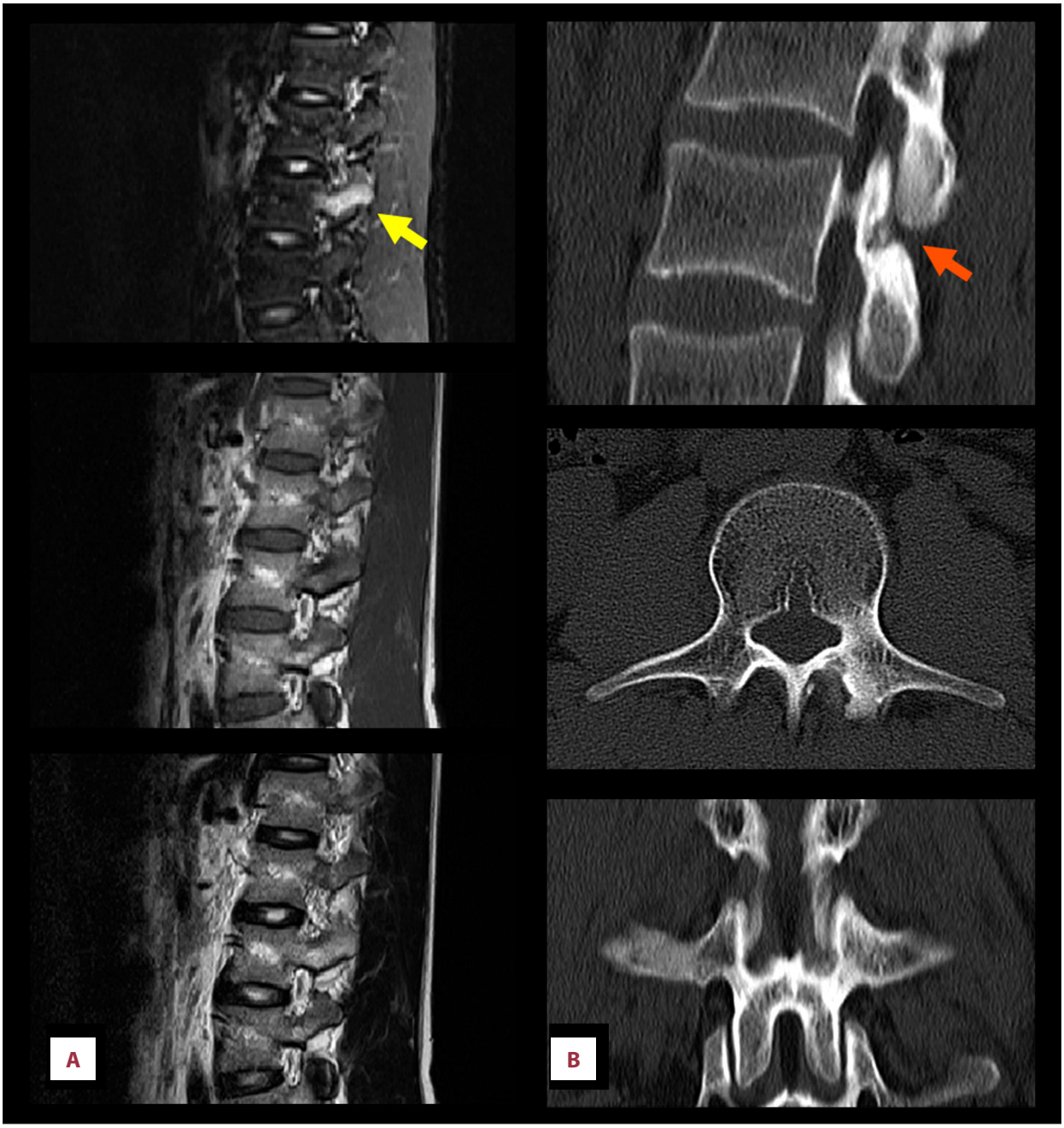

Figure 2. Osteoid osteoma of the lumbar spine. Bone marrow edema with high signal intensity at the pedicle on a fat-suppressed T2-weighted image (yellow arrow) (**A-top**), while normal on T1-weighted (**A-middle**) and T2-weighted (**A-bottom**) images without fat-suppression. CT with a 5-mm slice thickness showing a small osteolytic lesion with surrounding osteosclerosis at the pars interarticularis (orange arrow) (**B**) (top; sagittal, middle; axial, bottom; coronal). The lesion is less obvious on the axial (**B-middle**) and coronal (**B-bottom**) views.

six months after the onset. To determine the possible cause of the pain, analysis of MRI and CT images were performed at the same hospital. Based upon the MRI and CT findings, the patient was referred to our institute with a diagnosis of lumbar spondylolysis.

He had no previous history of low back pain besides his current history. No trauma related to the lumbar pain existed. He did not participate in any sports activity. No spinal developmental disorder, such as scoliosis, had been detected, and no bone-associated systematic disease had been detected, including endocrine disorder. He was a laborer and worked as an electrical

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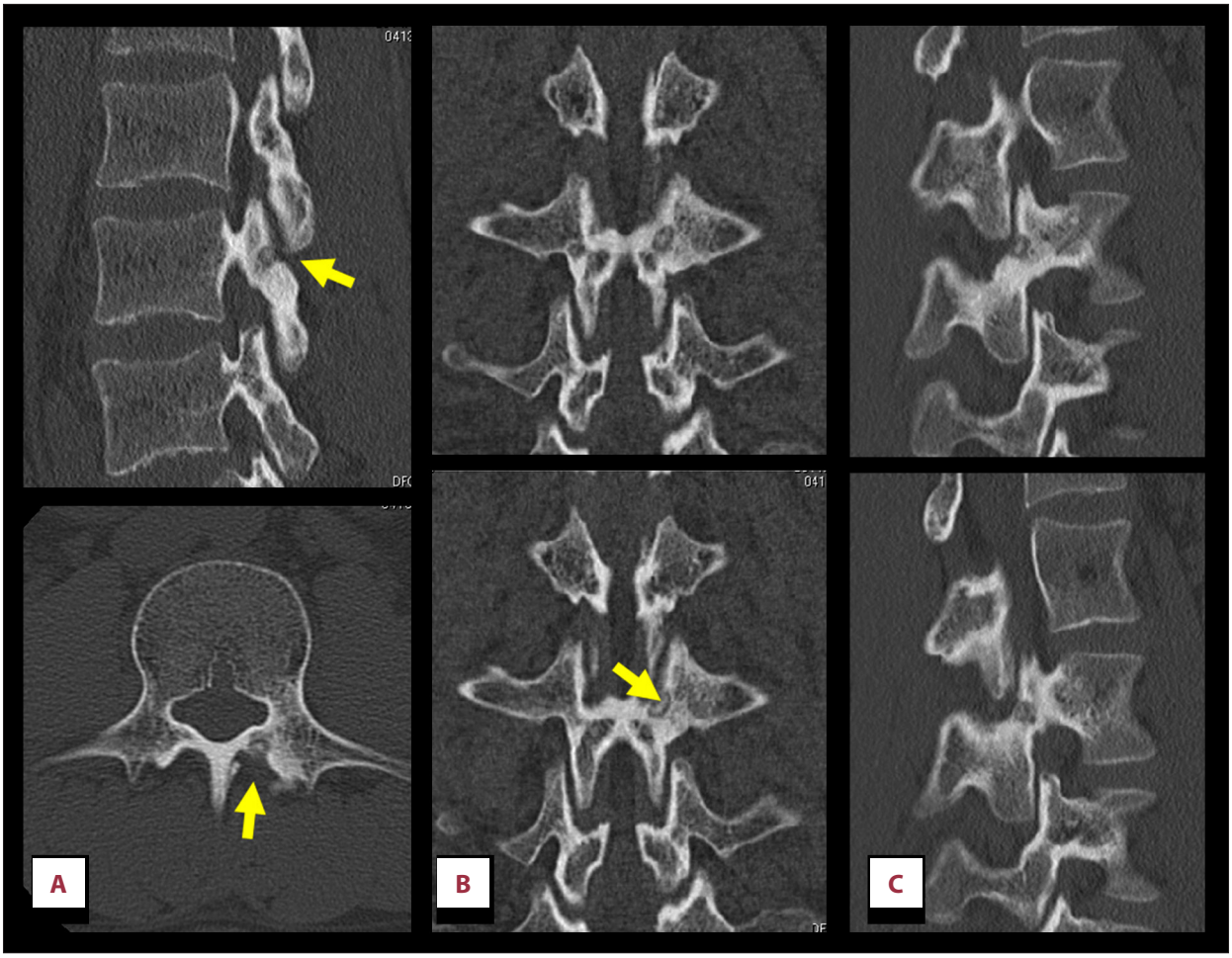

Figure 3. CT with a 1-mm slice thickness clearly showing a nidus of osteolysis with internal ossification (yellow arrows) at sagittal (**A-top**), axial (**A-bottom**), coronal (**B**), and oblique (**C**) views. Top and bottom panels show different sections of coronal (**B**) and oblique (**C**) views.

construction engineer. He often carried materials heavier than 20 kg during his work. He was 178 cm tall and weighed 88 kg. On physical examination, there was a slight tenderness over his left paravertebral muscle. No local heat, swelling, or redness was seen. No sensory or motor disturbance including the local area, or radiculopathy were observed. The straight leg raising test, which is sciatic nerve stretch test, gave negative results. The deep tendon reflexes at both the Achilles tendons were equally normal. The lack of finding of radiculopathy suggested less possibility of lumbar disc herniation.

Plain radiographic imaging was apparently normal. Discontinuity or osteosclerosis at the pars interarticularis, which might lead to a diagnosis of lumbar spondylolysis, was not observed at the oblique view of the plain radiograph (Figure 1). Although a diagnosis of lumbar spondylolysis was apparently unsupported by plain radiography, considering his age and occupation, lumbar spondylolysis was the most likely diagnosis.

On MRI of the lumbar spine taken at the previous hospital, T1- and T2-weighted images showed no obvious abnormalities. However, fat-suppressed T2-weighted images revealed high signal intensity at the pars interarticularis of the lamina and at the pedicle on the left side of the third lumbar (L3) vertebrae, but not at the surrounding soft tissue (Figure 2A). The high signal intensity on fat-suppressed T2-weighted images was compatible with bone edema because of spondylolysis, as found in a fracture. On the MRI, infection lesions, such as epidural abscess were not observed. CT showed osteosclerosis at the pars interarticularis of the lamina. The only surrounding osteosclerosis at the pars interarticularis was compatible with a very early stage of spondylolysis. However, a small osteolytic lesion (2 mm nidus) was found in reconstructed sagittal CT views at our institute (Figure 2B). The osteolytic lesion was previously overlooked on axial and coronal views, although the lesion was confirmed retrospectively.

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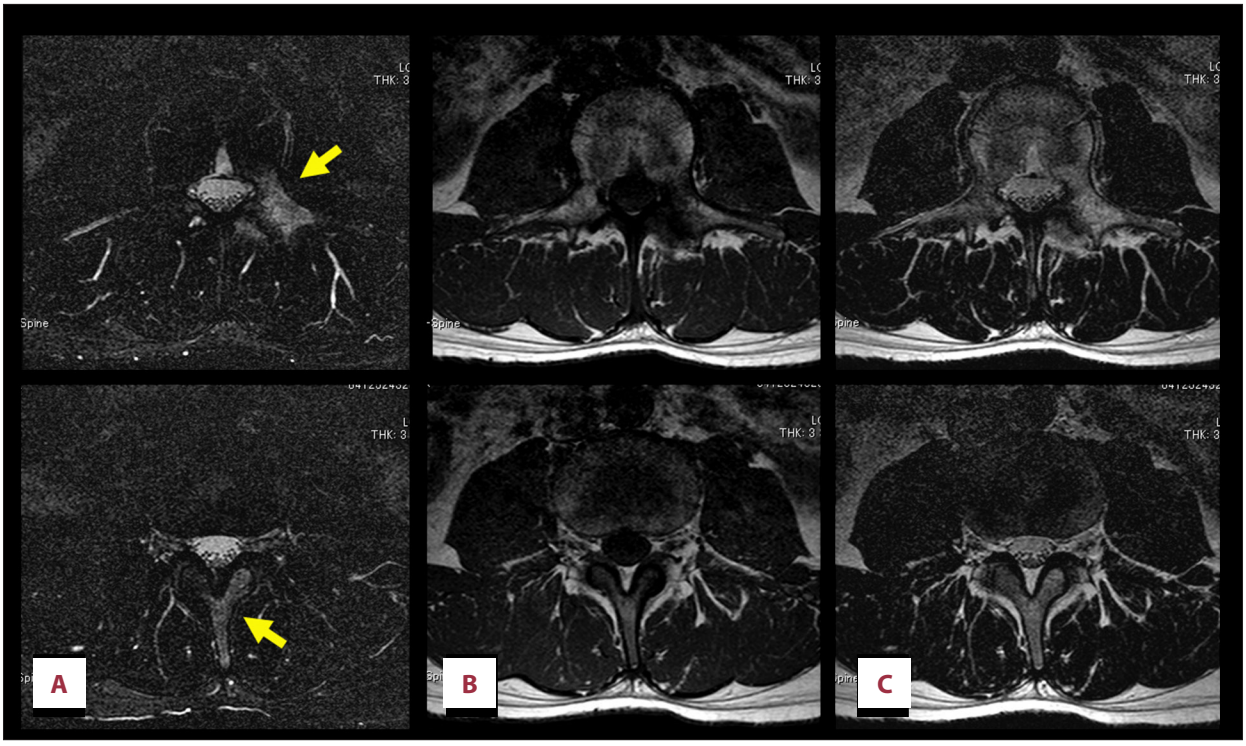

Figure 4. High signal intensity is seen at the pedicle (top) to spinal process (bottom) on fat-suppressed T2-weighted images (yellow arrows) (**A**). There is no obvious abnormal signal intensity on T1-weighted (**B**) and T2-weighted (**C**) images without fat-suppression.

Thin-sliced CT (1 mm slices) was performed at our institute. The CT depicted a round shaped osteolysis with ossification inside, and surrounding osteosclerosis (Figure 3). This finding led to a diagnosis of osteoid osteoma. Because the axial views of the previous MRI were only assessed at the disc level and not at the vertebral body, MRI was performed again to include consequent axial views including the whole vertebrae. Fat-suppressed T2-weighted images showed bone marrow edema extending from the pedicle to the spinal process, which is unusual for spondylolysis (Figure 4). Taking into consideration the persistent pain and imaging findings, we made a diagnosis of osteoid osteoma at the pars interarticularis.

The osteolytic lesion was surgically excised. With the patient under general anesthesia, and in a prone position, we used a posterior midline approach to expose the L3 lamina on the left side. Thinned cortex and a soft lesion under the cortex were confirmed. The lesion was curetted, and the surface of the surrounding osteosclerosis was removed with a high-speed drill. Histology of the curetted tissue failed to include a typical nidus, but included reactive bone formation, possibly associated with an osteoid osteoma. There was no malignant tissue, and the lack of atypia in the formed bone tissue indicated no possibility of osteosarcoma.

The preoperative pain disappeared after the surgery, except for mild pain at the surgical site. No complication associated with surgery occurred, and no neurological defect or muscle weakness was observed. Considering his clinical course, the images, and the histology, a diagnosis of osteoid osteoma was suggested. A plain radiograph obtained one month after the operation showed no abnormality (data not shown). Three months after the operation, his surgical site pain had disappeared. At one year after the operation, regular follow-up was ended because the patient worked without any pain. However, he was advised to visit the institute if the same lumbar pain reoccurred.

Discussion

In the current article, we described a patient who presented with a six-month history of low back pain responsive to NSAIDs with a concern for lumbar spondylolysis. However, subsequent workup concluded a diagnosis of a pars osteoid osteoma. Surgical removal of the benign lesion completely resolved the patient's pain without postoperative complications.

The pain of osteoid osteoma can be persistent, and is typically worse at night. However, pain at night has been reported to occur in only about 30% of patients with an osteoid

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osteoma [13]. Osteoid osteoma without pain has been reported at a prevalence of 1.6% [14]. NSAIDs are effective for the pain of osteoid osteoma [5]. However, NSAIDs are also effective for nonspecific low back pain. Therefore, effectiveness of NSAIDs is not sufficient to diagnose osteoid osteoma without supportive imaging.

In patients with an osteoid osteoma, plain radiographs often fail to depict the nidus as an osteolytic lesion because of the small size of the nidus. By contrast, CT is often useful to show the nidus with varying degrees of perinidal sclerosis [4]. However, previously normal-sliced CT failed to show any abnormality, and thin-sliced CT was recommended to show the nidus [15]. In the present case, CT with 1 mm slices showed the nidus with a clear, round osteolytic lesion ossification inside, leading to a diagnosis of osteoid osteoma, although the nidus had been overlooked in CT with 5 mm slices.

Lumbar spondylolysis is a stress fracture of the pars interarticularis, and is classified into three stages: early, progressive, and terminal [16]. An early defect was defined as a fissure in the pars. At the progressive stage, the defect is narrow, but the edge is round. The terminal stage is characterized by a wide defect with osteosclerosis [17]. Early stage lumbar spondylolysis is difficult to diagnose using plain radiography. Because of the anatomical complexity of vertebrae, CT is useful for depicting lesions of spondylolysis, even when a plain radiograph is normal. However, even using CT, the fracture lines are sometimes faint and therefore lumbar spondylolysis may be difficult to diagnose at a very early stage spondylolysis [12]. Osteosclerosis is a finding for early stage spondylolysis, because the pathogenesis of spondylolysis is considered to be a stress fracture [10,11]. In the present case of osteoid osteoma, the findings of osteosclerosis associated with osteoid osteoma at the pars interarticularis at the initial CT were misdiagnosed to be a very early stage spondylolysis.

At early-stage lumbar spondylolysis, high signal intensity on fat-suppressed T2-weighted images is able to depict spondylolysis [12]. The typical MRI findings of early-stage lumbar spondylolysis are high signal intensity on fat-suppressed T2-weighted images in the pedicle or pars, or both [12]. By contrast, MRI findings of the nidus are typically low signal intensity on T1- and T2-weighted images with surrounding bone marrow edema [18]. The nidus could not be detected because of its small size and the low signal intensity, which is often masked by the associated perilesional edema surrounding the nidus [18]. In the present osteoid osteoma at the pars interarticularis, fat-suppressed T2-weighted images of the MRI depicted bone marrow edema extending from the pedicle to the spinal process, which is an unusual finding for spondylolysis.

Lumbar disc herniation is one of the most common causes of low back pain in adolescents. The one reason for failing to detect the extensive edema of osteoid osteoma at the initial MRI was that the axial assessment was only performed at the disc level, not at the middle vertebral level, because the axial MRI views were used to assess the lumbar discs. A previous case report indicated that osteoid osteoma in the lumbar spine could cause radiculopathy and low back pain because of the inflammation caused by the osteoid osteoma [19]. In the present case, lumbar disc herniation was not a differential diagnosis, because of the lack of any radiculopathy and no findings of herniation on MRI. If there were findings of disc bulging and herniation, hiding the cause of radiculopathy, osteoid osteoma might have been overlooked, and its diagnosis would have been delayed.

As a limitation of the present diagnosis, the histology failed to show a typical nidus, but showed only reactive bone formation, possibly associated with an osteoid osteoma. In a previous report, as an explanation of this failure, it had been suggested that the nidus was removed by the surgeon but was not recognized or sent for histological examination, and that perhaps a nidus was present [20]. In our present case, because the size of the nidus was small at 2 mm diameter, the nidus was lost during pathohistological procedures. Nevertheless, the clinical course and the images were typical of osteoid osteoma.

Conclusions

In the present report, a case of osteoid osteoma occurring at the pars interarticularis of the lumbar spine was described. At a very early stage spondylolysis, the defects may be faint. It was challenging to make a diagnosis of osteoid osteoma based on MRI and CT findings because of the similarity to very early stage spondylolysis except for the existence of nidus in osteoid osteoma. The possibility of osteoid osteoma should be kept in mind when examining young patients who have low back pain. Furthermore, examination of thin-sliced CT and MRI of the entire vertebrae is important when spondylolysis is not seen, not only for diagnostic certainty, but also to exclude a diagnosis of osteoid osteoma.

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