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A case of Osteomalacia Mimicking Spondyloarthritis

Spondiloartriti Taklit Eden Bir Osteomalazi Olgusu

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Introduction

Osteomalacia (OM) is a metabolic bone disease characterized by inadequate mineralization of newly formed bone matrix called osteoid. The main problem is the lack of available calcium or phosphorus or both. The pathognomonic finding of OM is Looser's zones or pseudofractures, a thin transverse band of rarefaction, which can be seen on roentgenograms (1). The diagnosis of OM is often delayed, since it can be easily mixed with other pathologies, such as fibromyalgia, polymyalgia rheumatica, hypothyroidism, inflammatory myopathies, multiple myeloma, metastatic bone disease, rheumatoid arthritis, and ankylosing spondylitis (AS) (2). In this report, we describe a patient with OM who was misdiagnosed as spondyloarthritis (SpA) and treated with an anti-tumor necrosis factor (anti-TNF) agent but relieved only after receiving vitamin D and calcium.

Case Report

A 26-year-old woman was admitted with difficulty in walking and pain in her back and hips. Her complaints had started 4 years ago after she fell down the stairs. She had been diagnosed with lumbar disc hernia and received different medical treatments for 2 years. Later on, magnetic resonance imaging (MRI) of the hip revealed pathological signal changes in her left sacroiliac joint (acute sacroiliitis?) and bilaterally in the caput femoris and an old fracture on the anterior side of her right sacrum. Then, she was referred to a rheumatology department with a prediagnosis of SpA. Since the MRI findings of the sacroiliac joint was also compatible with sacroiliitis, considering her low back pain which seemed to be inflammatory, she was diagnosed with SpA and started to receive indomethacin plus sulfhasalazine treatment. After a while, her pain did not relieve, thus, etanercept was added to the treatment. However, 3 months later, since she had no benefit, the treatment was quitted and she was referred to the physical therapy and rehabilitation department for consultation.

On physical examination, it was observed that she was walking slowly with difficulty in a duck-like waddle. Getting into sitting or laying position was very difficult and the movements of bilateral hip joints were restricted and painful. She had severe pain on palpation of the symphysis pubis.

Serum biochemical analysis revealed decreased calcium level of 8.1mg/dL (normal 8.4-10.5) and decreased phosphorus level of 2.3 mg/dL (normal 2.3-4.7). Alkaline phosphatase and parathormone levels were 28 IU/L (normal: 20-155) and 311 pg/mL (normal: 12-72), respectively. Serum 25-hydroxyvitamin D had decreased to 3.8 ug/L. All other routine hematology, serology (including ESR and CRP) and biochemistry tests were within normal limits. Human leukocyte antigen-B27 was also negative.

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Plain radiographs revealed symmetric fracture zones in both of her superior pubic rami and inner margins of the femoral neck (Figure 1). MRI of the sacroiliac joints was repeated to rule out concomitant sacroiliitis. This time, MRI of the sacroiliac joints was interpreted as normal. Plain radiographs of her feet, which were requested for her right foot pain, showed a fracture zone in her right fibula. Therefore, we ordered a bone scintigraphy in order to see if there were any more fractures. Bone scintigraphy revealed multiple areas of increased activity in the ribs along with the previous findings, all of which were found to be compatible with pseudofractures (Figure 2). Considering the fact that her urinary system functions were normal, she did not have any pathological finding in her gastrointestinal system, and she had highly traditional outdoor clothing which prevented the action of sunlight on the skin, we made a diagnosis of OM due to vitamin D deficiency. Then, we started treatment with vitamin D3, 300.000 IU per week intramuscularly for 3 weeks, 1 µg alpha calcidiol and 1000 mg elemental calcium plus 880 IU vitamin D3 per day orally. She was also given analgesic and non-steroidal anti-inflammatory drugs for pain management.

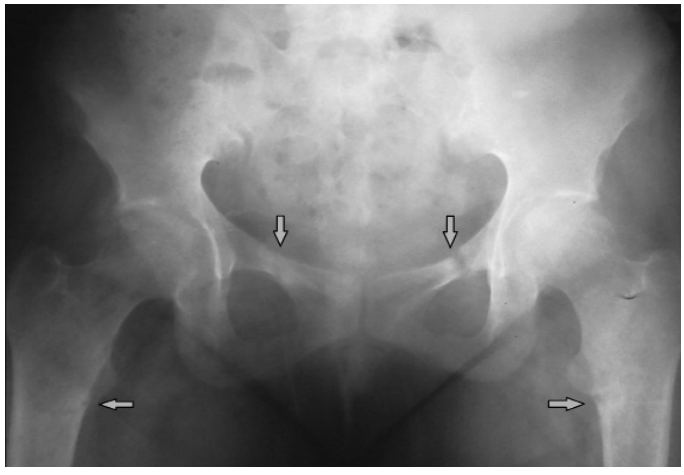

Figure 1. Radiograph of the patient showing symmetric fracture zones in both superior pubic rami and inner margins of the femoral neck.

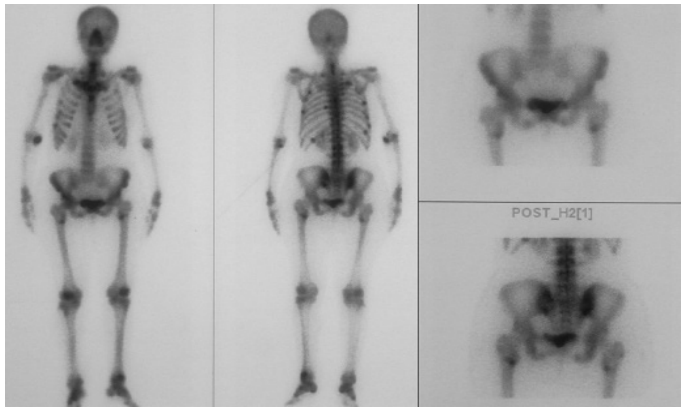

Figure 2. Bone scintigraphy revealed increased activity involvement in multiple ribs, both superior pubic rami and inner margins of the femoral neck that were compatible with pseudofractures.

During the second week of the treatment, there was a remarkable decrease in her complaints. After the pain subsided, by the third week of the treatment, range of motion and isometric strengthening exercises were given. By the third month, clinically and the sixth month, radiologically, a significant improvement could be seen. At the end of the fourteenth month, there was a progressing healing of the pseudofracture at the left superior pubic rami while the other areas had already recovered radiologically (Figure 3,4).

Discussion

OM is a relatively common but easily missed metabolic bone disease because of its nonspecific symptoms such as diffuse aches and pain in the joints and bones as well as proximal muscle weakness. The most common cause of OM is vitamin D deficiency, making it an easily treatable disease (2). Despite the abundant sunshine, vitamin D deficiency is common in Turkey even in the young population because of poor sunlight exposure and strict clothing style due to socio-cultural and religious reasons in some regions (3,4).

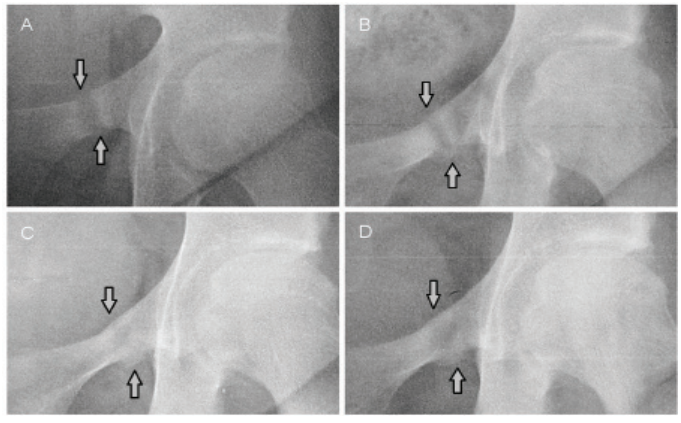

Figure 3. A, Fracture zone in the left superior pubic ramus obtained before the treatment. B, Follow-up radiograph obtained 6 months after the treatment. C, Follow-up radiograph obtained 10 months after the treatment. D, Follow-up radiograph obtained 14 months after the treatment.

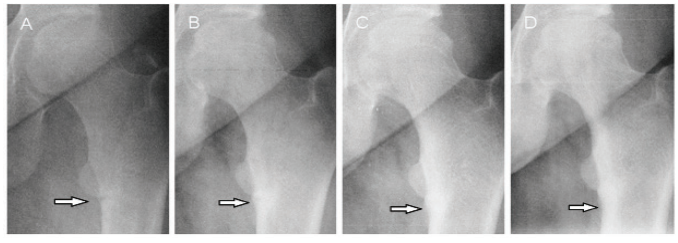

Figure 4. A, Fracture zone in the left femoral neck obtained before the treatment. B, Follow-up radiograph obtained 6 months after the treatment. C, Follow-up radiograph obtained 10 months after the treatment. D, Follow-up radiograph obtained 14 months after the treatment.

The classical radiologic lesion of OM, which is pathognomonic, is Looser's zone, also known as pseudofracture or Milkman's zone which are seen as multiple, bilateral, symmetric radiolucent lines placed in the cortex perpendicular to the long axis of the bones. These lesions representing cortical stress fractures filled with poorly mineralized callus, osteoid, and fibrous tissue, are common along the axillary margins of the scapula, the inner margin of the femoral neck, the proximal dorsal aspect of the ulna, the ribs, and the pubic and ischial rami (2).

Depending on the predominant clinical feature, OM can be confused with several disorders, one of which is AS. Even though it is not found to be common in the literature, in case of axial involvement of OM, fuzzy sacroiliac joint outlines, calcified enthesopathies and subchondral bone resorption can be seen as in AS (5-8).

Other than our case, in the literature, there are only a few cases of OM mimicking AS. One of them has been reported in the New Jersey series. In these series, among 22 patients who had the final diagnosis of biopsy-proven OM, only one of them had AS-like findings initially (1). Another case has been reported from our country. This was the case of a 39-year-old woman whose symptoms and physical findings had fulfilled the modified New York criteria for AS, while her radiologic and laboratory abnormalities brought the diagnosis of OM instead of AS (9). The problem that the author considered in that case was if the case was purely OM or presented together with AS. In the literature, there are also a few cases that had the final diagnosis of both AS and OM. The one thing common in almost all of these cases was accompanying hypophosphatemia (5,10-12).

Now that the new guidelines of the Assessment of Spondyloarthritis International Society are available, early diagnosis of axial SpA is possible. With these new criteria, in patients whose age of onset is less than 45 years and having back pain for at least 3 months, findings of sacroiliitis on MRI plus at least one SpA feature is enough to make the diagnosis (13). By this way, it is possible to identify the disease much earlier and it enables the use of anti-TNF agents at an early stage. However, these new criteria also necessitate the correct interpretation of the sacroiliac MRI before making the definite diagnosis of sacroiliitis. In our case, the misinterpretation of the MRI, together with inflammatory back pain and heel enthesitis had resulted in misdiagnosis of SpA and led to aggressive treatment protocols. Therefore, the differential diagnosis should be well done.

In conclusion, it is important to have a high index of suspicion for OM, which is a much more common and easily treatable condition, before making the definite diagnosis of SpA.

Conflict of Interest

Authors reported no conflicts of interest.

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