BILATERAL SACROILIAC MILKMAN FRACTURE AND SACROİLİİTİS: CASE REPORT

SAKROİLEİT VE BİLATERAL SAKROİLİAK MİLKMAN FRAKTÜRÜ: VAKA SUNUMU

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ABSTRACT

Osteomalaciais a metabolic disease, which is characterized with impaired bone mineralization, and which mimics other rheumatic diseases due to its clinical signs. Diffused pain, proximal muscle weakness, and paresthesia are among its clinical signs. In this case report, we presented the female patient, who applied to our outpatient clinic with low back pain, and who was detected to have bilateral sacroiliac milkman fracture due to osteomalacia in examinations previously performed with pre-diagnosis of spondyloarthritis.

Keywords: Osteomalacia, milkman fracture, sacroiliitis

ÖZET:

Osteomalazi kemikteki mineralizasyon bozukluğuyla seyreden, klinik belirtileri nedeni ile diğer romatizmal hastalıkları taklit eden metabolik bir hastalıktır. Klinik belirtileri arasında yaygın ağrı, proksimal kas güçsüzlüğü, parestezi vardır. Bu olguda polikliniğimize bel ağrısı ile başvuran, öncesinde spondiloartropati ön tanısı ile yapılan incelemelerde osteomalazi nedeni ile bilateral sakroiliak milkman fraktürü tespit edilen bayan hastayı sunduk.

Anahtar Kelimeler: Osteomalazi, milkman fraktürü, sakroileit

INTRODUCTION

Osteomalacia is a metabolic disease, characterized with increased non-calcified matrix amount due to impaired bone mineralization. Its most frequent cause is Vitamin D deficiency.Diffused bone pain, tetany in presence of hypocalcemia, paresthesia, convulsion, and muscle weakness might be observed as clinical signs.Lowornormal serum calcium(Ca),low or normal serum phosphorus (P),high parathyroid hormone (PTH), and low 25-hydroxy (OH)vitamin D3level are diagnostic factors. Milkmanfracture, height loss in vertebra, and framework signs are present in radiology(1).Osteomalacia is frequently misdiagnosed as other rheumatic diseases due to its signs. Some examples of these diseases

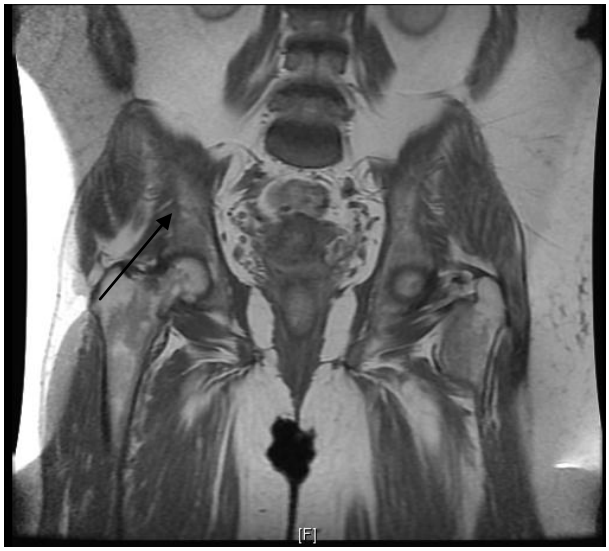
mimicked are fibromyalgia, hypothyroidism, metastatic bone diseases, polymyalgia rheumatica, inflammatory myopathy, multiple myeloma, rheumatoid arthritis and ankylosing spondylitis (AS)(2).In this article, we presented our case, who applied to the outpatient clinic with low back pain and morning stiffness, who was pre-diagnosed with sacroiliitis and then who was detected to have bilateral sacroiliac milkman fracture in the radiological examination performed.

CASE

39-year female patient applied with complaints of low back and right hip pain, not being able to walk for the last one year, morning stiffness for 1-2 hours and nocturnal pain.Our patient, who was a housewife, had a clothing style with only face and hands uncovered. In her physical examination, she was transferred with wheel chair, and she had no balance to stand up with no support. In patient's posture assessment, it was observed that her body had forward flexion and increased lordosis. Hip joint range of movement was painfully open, her Visual Analog Scale (VAS) for pain was 10 and her motor, sensory and reflex examinations were normal.Bilaterallaseque test was negative, sacroiliac compression and stretching test was positive at right. In her blood tests, serum calcium level was 9.3(N:8.4-10.6) mg/dl, serum Plevel was 1.8 (N:2.3-4.7)mg/dL,alkaline phosphatase (ALP) was 295 (N:30-120) IU/L,PTH was 406(N:12-88)pg/ml,25OH D3 was 6.5(N:20-120)µg/L and spot urine calcium was 2 (N:12-20) mg/dl and24-hour urine calcium was 24 (N:100-300)mg/day (urine volume was 1200ml).All other hematological, biochemical and serologic test results were within normal limits. In patient's pelvis radiography performed, irregularity was detected at bilateral sacroiliac joint. In sacroiliac MRG, bilateralsacroiliacmilkmanfracture was detected (Figure 1-2).In DEXAgraph, lumbal Total T score was -3.0,and femurneckTscore was -3.7.After the detection of patient's osteomalacia, the loading dose of vitamin D 100000 IU/week for 8 weeks was initiated and Calcium 1200mg+ vitamin D 800IU per day were given as maintenance.6 weeks after the treatment, VAS was 6,serum ALP was 266IU/L, PTH was 154.1,P was 3mg/dl,25-OH Vitamin D3 was 26,23 µ/L. Patient was ambulated with crutch, improving from wheelchair use. In month 3, serum ALP was 314 IU/L,PTH was 73pg/ml,P was 3mg/dl,25-OH D was 29,5 µ/Land patient's VAS regressed to 4.In 3-month follow-up, the patient was ambulated with walking stick and she could climb up and come down the stairs.

DISCUSSION

Diffuse body pain, sacroiliac joint pathologies, calcification at entheses area and presence of spinal pathologies, which are observed in osteomalacia, cause the disease to be misdiagnosed especially as spondyloarthropathies(3-7). Cases presenting spinal and sacroiliac joint involvement and being diagnosed with axial osteomalacia are available in the literature. Axial osteomalacia is a rare pathology of unknown etiology. It has been initially defined in 1961 by Frame et al. They have reported dense, rough and trabecular pattern in radiographic examination of axial skeleton. They have detected that the most apparent involvement was at cervical spine and that involvement was also present at lumbar spine, pelvis and costas. They have found the other radiographic examination of the skeleton normal and bone biopsy samples indicated histological osteomalacia(6).



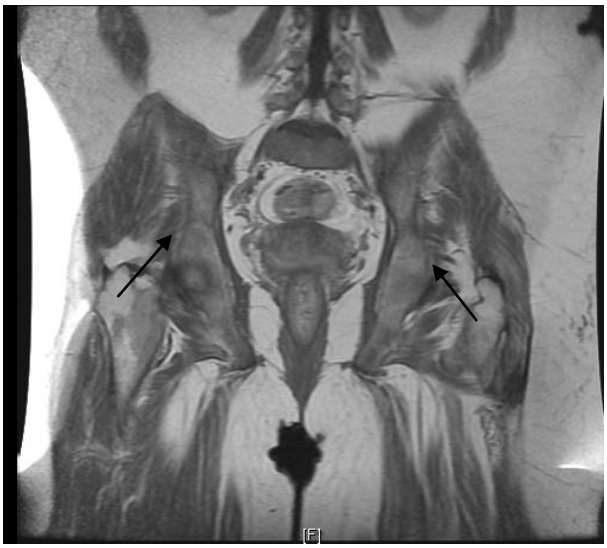


Figure 1:Fracture line at right iliac wingFigure 2: Fracture line at bilateral iliac wings

Even though osseous structure is completely affected in osteomalacia, radiographic diagnosis is difficult.Decreased bone density, rough trabecular pattern, corticalirregularity, pseudo-fractures, and deformities are radiological findings of osteomalacia.Pseudo-fractures, defined initially by Looser in 1920 and confirmed by Milkman in 1934, are the most significant radiological finding of the disease.The most frequently observed areas are femurneck, pubicand ischialramus,ribs, and scapula.In osteomalacia diagnosis, it is considered that mid-section of sacroiliac joint is dominantly affected due to insufficient demineralization. Gaucherhas shown the association between the bone pains of a 20-year old Fanconisyndrome patient and early sacroiliac deterioration(8).

In our case, firstly spondyloarthropathy was considered since sacroiliaccompression and stretching test was positive at right, morning stiffness lasted for 1-2 hours, low back and right hip pain were present, nocturnal pains were present, and patient's age was 39 years.Since patient's ALP value was high, vitamin D was requested. Upon observation of irregularity at bilateral sacroiliac joint in pelvis A-P graph, sacroiliac MR was performed. Due to assessment of milkman fracture on bilateral sacroiliac surface in sacroiliacMR, SpAdiagnosis was disregarded. Şahinet al. have detected sacroiliitis in 2 women with osteomalacia(9). Akkuşet al. have treated a patient having positive sacroiliac tests with osteomalacia diagnosis(10).Nevertheless, GaucherA et al. have reported that pseudo-sacroiliitis may be observed in osteomalacia(8).However, bilateralsacroiliac milkmanfracture is not available in the literature. SarıyıldızM A et al. have published unilateralmilkmanfracture(11).Onur O et al. have defined a subchondral sacroiliac bone fracture presented as adolescent sacroiliitis(12). Mısırlıoğluet al. have started to treat a patient with pre-diagnosis of spondyloarthropathy after the detection of left sacroiliitis, however, since there was no clinical relief, osteomalacia was diagnosed with the results of tests performed, and treatment was initiated, and they have observed a very fast improvement(13).

Our patient's Dexa graph performed was leading to osteoporosis diagnosis. Her lumbar TotalT score was -3.0. In the study performed by Reginato AJ et al., concomitant presence of osteoporosis with osteomalacia has been indicated through diagnosis of 6 patients by

biopsy(14).There are also studies which indicate that hypovitaminosis D cause especially lower lumbar spine Z scores compared to controls(15-18).Our case had osteoporosis in consistency with these studies. Nevertheless in a 33-year female AS patient, AS was associated with osteomalacia and post-pregnancy spinal osteoporosis at postpartum month 3(19).Very few cases of concomitant osteomalacia and ankylosing spondylitis are available in the literature. Hypophosphatemia was concomitantly present in all these cases(6,20,21). In our case as well, hypophosphatemia, which was existing at baseline, recovered with vitamin D treatment, and in addition to this, clinical findings suggestive of spondyloarthropathy also improved. In connection with this, we concluded that sacroiliac fractures in osteomalacia might be clinically suggestive of sacroiliitis.

In conclusion, if clinical findings for sacroiliac joint involvement are present in osteomalacia cases, possible sacroiliac fracture should also be considered. Interestingly, bilateral sacroiliac milkman fracture may also be observed as it has been the same in our case.

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