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Functional Parathyroid Adenoma Diagnosed After Stress Fractures

Stres Kırıkları Sonrasında Tanı Alan Fonksiyonel Paratiroid Adenomu

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Clinical findings and radiographs may be disleading or indefinite in patients with insufficiency fractures. Herein, we present a patient with low back pain increasing with activity and morning stiffness, which initially were thought to be due to a spondyloarthropathy. Magnetic resonance imaging (MRI) revealed insufficiency fractures in the sacrum and iliac wings bilaterally. Finally, laboratory and imaging findings depicted a parathyroid adenoma causing hyperparathyroidism and the adenoma was removed surgically.

Key Words: ***stress fractures, parathyroid adenoma***

Yetmezlik kırıklarında klinik ve radyografik bulgular her zaman belirgin olmayabilir ya da doktoru yanlış yönlendirebilir. Biz burada hareketle artan bel ağrısı ve sabah tutukluğu nedeni ile başvuran ve ilk etapta spondiloartropati lehine değerlendirilen bir hastayı sunuyoruz. Yapılan manyetik rezonans görüntüleme (MRG) ile sakrum ve bilateral iliak kanatta yetmezlik kırıkları saptanan hastada sonuçta laboratuvar ve görüntüleme bulguları ışığında hiperparatiroidizme neden olan bir paratiroid adenomu tesbit edildi ve cerrahi olarak çıkarıldı.

Anahtar Kelimeler: ***stres kırıkları, paratiroid adenomu***

Insufficieny fractures which are a subgroup of stress fractures, occur by normal or physiologic stress uploaded on an abnormally weakened bone. Hyperparathyroidism is one of the risk factors of insufficiency fractures besides diseases like postmenopausal osteopenia, rheumatoid arthritis, osteomalacia, and fibrous dysplasia. Clinical findings and radiographs may not always be adequate for the diagnosis of sacral insufficiency fractures. The patient we present here had low back pain increasing with activity and morning stiffness which in the first place were thought to be due to a spondyloarthropathy. Magnetic resonance imaging (MRI) showed the insufficiency fractures in the sacrum and iliac wings. The laboratuary and imaging findings depicted a parathyroid adenoma causing hyperparathyroidism and the adenoma was removed surgically.

Case

A 50-year-old woman was admitted to the rheumatology department with a 3 months history of low back pain which increased with exercise and morning stiffness of 15 minutes. No signs of inflammatory bowel disease or uveitis were present. In the physical examination there was no neurologic deficit but the range of motion of the lumbosacral joint was impaired due to severe pain. Besides routine hematological and biochemical analyses immunological markers, HLA B27 antigen and protein electrophoresis were also performed in order to eliminate a spondyloarthropathy. The laboratory findings included an increase in alkaline phosphatase level (438U/l) but the immunological markers and protein electrophore-

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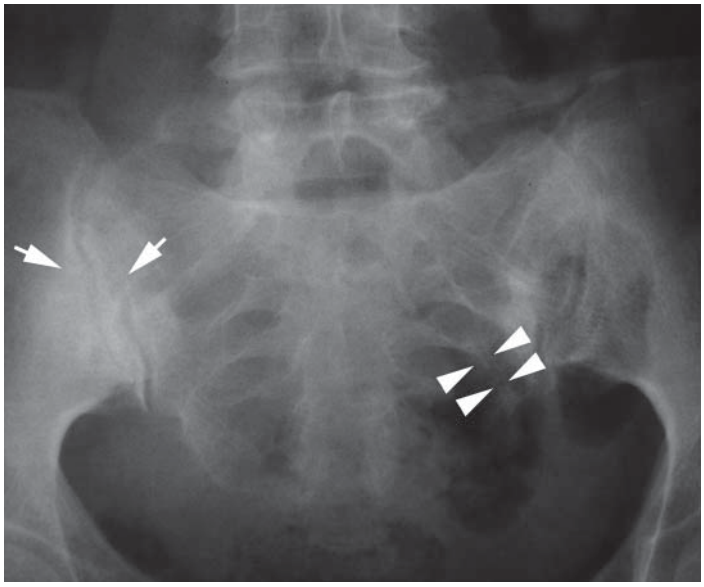

Figure 1. Plain pelvic radiogram showing sclerosis on the right sacroiliac joint (arrows). In the retrospective evaluation of the radiogram, an indistinct linear fissure on the left sacral wing was also noticed (arrow heads).

sis were normal and HLA B27 antigen was negative. There was marked sclerosis of the right sacroiliac joint on the plain radiographs of the pelvis (Figure 1). Sacroiliac MRI was performed to reveal possible sacroiliitis. MRI showed a linear low signal intensity area, mostly evident on T1-weighted images on the left sacral wing (Figure 2a) with associated bone marrow edema on the left sacral wing and iliac bone on FSEIR images (Figure 2b) which were found to be concordant with stress fractures. MRI also revealed low signal intensity areas without accompanying bone marrow edema on the bilateral iliac wings (Figure 2a,b) and these were consistent of with healed old fractures. In the retrospective evaluation of the plain pelvic radiographs, a linear fissure on the left sacral wing was also noticed (Fig.1). In the further biochemical analyses which were performed to evaluate the high alkaline phosphatase level, there was also an increase in the serum calcitonin (15.9pg/ml), osteocalcin (44.7 ng/ml) and parathormone (232 pg/ml) levels and a decrease in 25-Dihydroxy vitamin D (6mg/L). Serum calcium level was within the normal limits (9.2 mg/dl). The patient was presumed to have secondary hyperparathyroidism due to serum vitamine D insufficiency and depot vitamine D and Calcimax® treatment was administered. Control laboratory analyses in a four month period revealed an improvement in the serum parathormone and alkaline phosphatase levels although they were not within the normal limits. The serum calcium level had also increased slightly over the normal limits. Ultrasonographic and scintigraphic examinations revealed a parathyroid adenoma which was removed surgically. No postoperative complications occured. The final diagnosis was parathyroid adenoma and primary hyperparathyroidism. In a one year follow-up the patient was symptoms free, and her biochemical analyses were within normal limits.

Discussion

Primary hyperparathyroidism refers to the excessive production of parathormone which leads to increased resorption of bone, and usually causes hypercalcemia. In 80-85% of the patients, primary hyperparathyroidism is due to benign parathyroid adenomas. Hyperplasia of the parathyroid glands, and rarely parathyroid carcinoma are other causes of primary hyperparathyroidism. Patients may be asymptomatic or may have nonspecific symptoms including muscle weakness, myalgia, thirst, polyuria, etc. Diagnosis is usually made by biochemical screening showing the increased levels of parathormon which leads to hypercalcemia, hypophosphatemia, and hypercalciuria. Sestamibi scintigraphy scan, single photon emission computed tomography imaging, and/or neck ultrasound would help the surgeon to demonstrate the presence and loca-

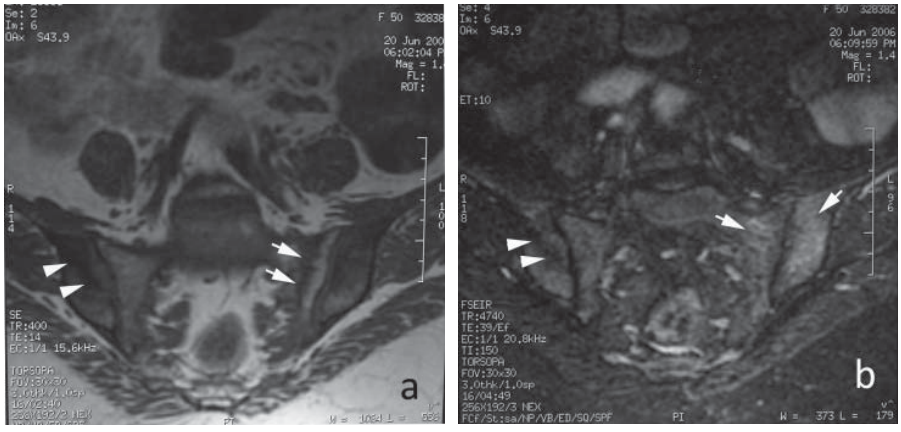

Figure 2. Coronal T1-weighted MR image shows a linear low signal intensity area (a, arrows), with associated bone marrow edema on the left sacral wing and iliac bone on coronal FSEIR image (b, arrows), which were found to be concordant with stress fractures. There is also a low signal intensity line without accompanying bone marrow edema on the right iliac wing (arrow heads on a and b) consistent of with healed old fracture.

tion of the parathyroid adenoma or the abnormal gland before surgery (1,2).

It should be kept in mind that an incidentally diagnosed insufficiency fracture may be an outcome of undiagnosed endocrinological disorders as well as other benign or malignant diseases. Postmenopausal osteopenia, prolonged corticosteroid treatment, pelvic irradiation, Paget disease, osteopetrosis, metabolic bone diseases like osteomalacia and hyperparathyroidism may be causes of insufficiency fractures (3). Multipl fractures may rarely be the onset of primary hyperparathyroidism and they usually represent late presentation of the disease (2). Chronic vitamin D deficiency as seen in our patient, is reported to be a risk factor for aggrevating the parathyroid adenoma growth, parathormone secretion and increased bone turnover (4,5).

The sacrum and iliac bones are the major weight-bearing structures of the body. Pelvic ring is the main site of insufficiency fractures and sacrum is mostly involved (6,7). Sacral fractures frequently occur in elderly women and the most common risk factor is osteoporosis. Low back pain with or without associated groin, hip or buttock pain is usually the leading clinical symptom. Physical examination and plain radiographs are indefinite or misleading in the vast majority of the cases. Most of the patients do not recall a major trauma and many of them are diagnosed incidentally by computed tomography (CT), MRI, skeletal scintigraphy or positron emission tomography (PET) studies performed for other reasons (8). The patient we present here also had unsuspected fractures which were diagnosed definitely by MRI, and there was no need for further radiological examination.

The sensitivity of the plain films has been reported as 34-37%. On the contrary, scintigraphy has a higher sensitivity (92.6%-97.3%) in the relevant literature and it sufficiently shows the sacral fracture (7).

The typical CT appearance of a stress fracture is a focal callus formation and endosteal thickening around a fracture line (8). CT scan should not be the initial examination technique and must be saved for the patients whose MRI imaging does not demonstrate the fracture line (7,9).

As MRI is widely being used for the patients with hip pain radiologists must be aware of the MR appearance of sacral stress fractures. The fracture line is typically hypointense on both T1- and T2-weighted images and in most of the cases there is a area of high signal intensity surrounding the fissure on T2-weighted sequences. This hyperintense area resembles inflammation or bone marrow edema seen in acute fracture (6).

In conclusion; MRI is a highly sensitive and spesific technique in the diagnosis of stress fractures. When multipl insufficiency fractures are detected, primary hyperparathyroidism should be remembered in the differrential diagnosis, and serum biochemical screening and neck examination are important investigations for detecting the underlying parathyroid adenoma.

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