Case Report

Bilateral Femoral Neck Fatigue Fracture due to Osteomalacia Secondary to Celiac Disease: Report of Three Cases

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Abstract

Bilateral non traumatic femoral neck fatigue fracture is a rare condition usually occurring secondary to medical conditions such as pregnancy, pelvic irradiation, corticosteroid exposure, chronic renal failure and osteomalacia. In this report, we present three young female patients with bilateral femoral neck fracture secondary to osteomalacia. The underlying cause of osteomalacia was Celiac disease in all patients. The patients were treated with closed reduction and internal fixation with cannulated lag screws. They were free of pain and full weight bearing was achieved at three months. There were no complications, avascular necrosis and nonunion during the follow up period. In patients with bone pain, non traumatic fractures and muscle weakness, osteomalacia should be kept in mind and proper diagnostic work-up should be performed to identify the underlying cause of osteomalacia such as celiac disease.

Keywords: Bilateral, celiac disease, femoral neck fracture,

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Introduction

Bilateral non-traumatic femoral neck fracture is a very rare condition that usually occurs secondary to medical conditions such as pregnancy, pelvic irradiation, corticosteroid exposure, chronic renal failure and osteomalacia.[1]

Osteomalacia is a disorder of mineralization of newly synthesized organic matrix caused by low levels of vitamin D. In severe cases, bone pain and weakness may cause the patient to be bedridden.[2]

Celiac disease has long been known to cause metabolic bone disease, usually osteomalacia secondary to calcium and vitamin D malabsorption.[3]

We report three cases of surgically treated bilateral non traumatic femoral neck fracture secondary to osteomalacia caused by celiac disease.

Case Reports

Case 1

A 30-year-old woman was referred to our clinic due to bilateral hip pain continuing for 4 months. She was diagnosed with celiac disease with histopathological confirmation during diagnostic work up of iron deficiency anemia three months ago. She described no trauma or fall. Physical examination of the lower limb revealed pain with hip movements. On the plain radiographs of both hips, there was no gross abnormal finding but bilateral femoral neck fractures were diagnosed with MRI. Her laboratory studies showed increased alkaline phosphatase (ALP) activity 305 U/L (reference range: 35–105 U/L), hypocalcemia (7.9 mg/dL), and hypophosphatemia (1.5 mg/dL). The 25-hydroxy-vitamin D [25(OH)D] level was 9.4 ng/mL (<20 ng/mL is defined as deficiency) and parathormone (PTH) level was increased to 152 pg/mL (reference range: 15–65 pg/mL) reflecting osteomalacia. Low bone mineral density (BMD) was found in dual-energy X-ray absorptiometry DXA (lomber: 0.52 g/cm^2; T score, -3.45; Z score, -3.35; left hip total: 0.58 g/cm^2; T score, -3.4; Z score, -3).

Case 2

A 35-year-old woman complained of progressive hip pain since one year ago. In the last 3 months, she was almost unable to walk. There was no trauma or fall. Physical examination of the lower limb revealed pain with hip movements. Although the plain X-ray of the hip was normal, the MRI of the hip showed bilateral femoral neck fractures (Figures 1 and 2). Her laboratory studies showed iron deficiency anemia, increased ALP activity 305 U/L, hypocalcemia (7.9 mg/dL), and hypophosphatemia (1.2 mg/dL). The 25(OH)D level was 5.1 ng/mL and PTH level was increased to 114.5 pg/mL reflecting osteomalacia. Upper gastrointestinal system (GIS) endoscopy was performed with suspicion of celiac disease because of iron and vitamin D deficiency. Then, CD was diagnosed histopathologically. Low BMD was found in DXA (lomber: 0.66 g/cm^2; T score, -3.5; Z score, -3.4; left hip total: 0.62 g/cm^2; T score, -2.6; Z score, -2.6).

Case 3

A 30-year-old woman was referred to our clinic due to bilateral hip and back pain for about 1 year. Recently, she was diagnosed with osteomalacia by an endocrinology clinic. As she also had iron deficiency anemia, an upper GIS endoscopy had been performed and celiac disease was diagnosed histopathologically. On her physical examination, the hip movements were restricted

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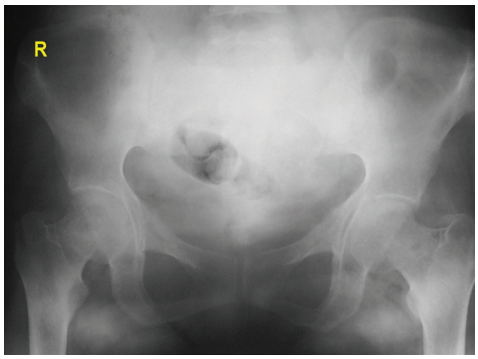

Figure 1. Pre-operative antero-posterior pelvic X-ray of the second case. **R:** Right side

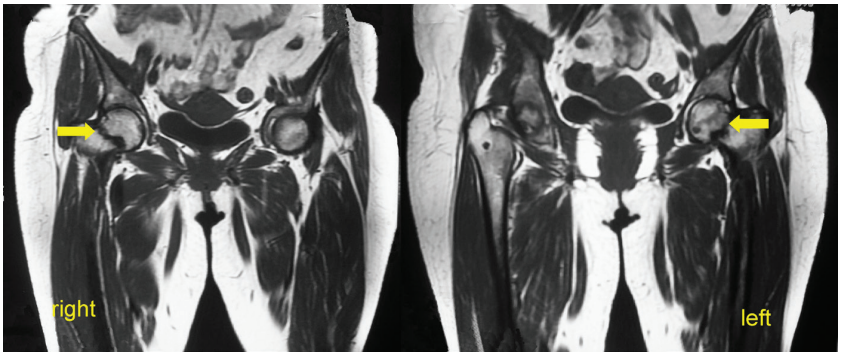

Figure 2. Pre-operative pelvic MRI of the second case. Arrows show the fracture line.

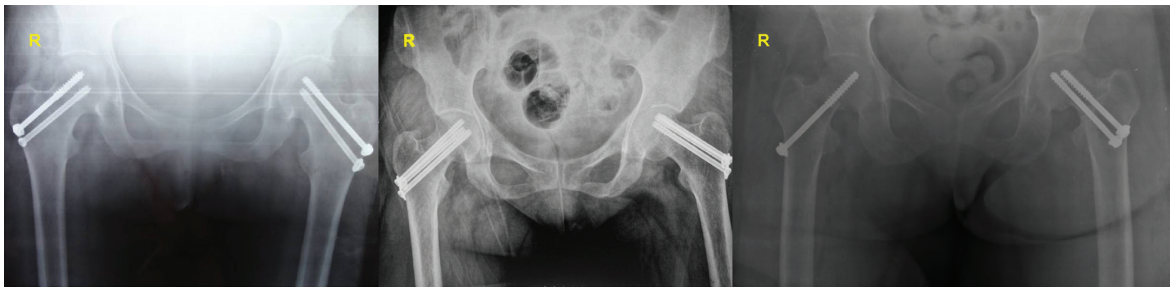

Figure 3. Early post-operative antero-posterior pelvic X-ray of three patients. **R:** Right side.

and painful. In addition, she had kyphosis. The patient was unable to walk alone. On plain radiographs of both hips, bilateral non-displaced femoral neck fractures were seen. The diagnosis was confirmed with computerized tomography (CT) images. At the same time, there were callus formations on ribs and angulation with posterior fusion of $T_{5\text{-}6}$ vertebrae on thorax CT images, which reflect previous fractures. She had no history of trauma. On laboratory evaluation, increased ALP activity 278 U/L, hypocalcemia (7.8 mg/dL), and hypophosphatemia (1.4 mg/dL) were found. The 25(OH)D level was 7.5 ng/mL and PTH level was increased to 109.1 pg/mL reflecting osteomalacia. Low BMD was found in DXA (lomber: 0.79 g/cm^2; T score, -3.2; Z score, -3.0; left hip total: 0.55 g/cm^2; T score, -3.6; Z score, -3.4).

Management and Follow-up

All patients underwent percutaneous screw fixation with cannulated lag screws under general anesthesia (Figure 3). There were not postoperative complications. The patients were not allowed to bear weight for six weeks. After 6 weeks, partial weight bearing was initiated with two crutches until three months. All patients were free of pain and full weight bearing was achieved at this time. Gluten free diet was initiated for all patients after celiac disease was confirmed, and adequate vitamin D and iron replacements were given.

Case 1 had four years, case 2 had 28 months and case 3 had 2 years of postoperative follow up. There were not any complications, avascular necrosis and non-union seen during the follow up periods.

Discussion

Osteomalacia is a disorder of mineralization of newly synthesized organic matrix, secondary to vitamin D deficiency in adults. The disorder involves only the bones. Vitamin D deficiency is usually the combined result of deficient sun exposure and decreased dietary intake or intestinal malabsorption. The disease can be also caused by metabolic defects in the vitamin D hormone system. The most common etiologic factors are nutritional and gastrointestinal disorders such as celiac disease, renal defects, oncogenic and drug induced osteomalacia. Parathormone levels

may be markedly increased and some of the bony changes of the disease are caused by secondary hyperparathyroidism.[2] In all our cases, PTH levels were increased secondary to vitamin D deficiency. The clinical expression of osteomalacia varies widely; in severe cases, bone pain and muscle weakness may cause the patient to be bedridden.[2]

Osteomalacia is a well-known extra intestinal manifestation of celiac disease. Celiac disease is an autoimmune small intestinal disease caused by gluten intolerance. Iron deficiency is one of the most common clinical manifestations of the disease due to malabsorption.[4] As all our patients had iron deficiency anemia and osteomalacia preoccupying malabsorbtion, upper GIS endoscopy was done for the second and third cases with suspicion of celiac disease, and the disease was diagnosed histopathologically. For the first case, a diagnosis of celiac disease had already been made. The symptoms of the disease may be silent and indistinct as seen in our second and third cases. The initial presentation of celiac disease in these two patients was bilateral femoral neck fracture. On reviewing the literature, there are some case reports of bilateral femoral neck fracture due to osteomalacia but only two of them were related to GIS etiology - celiac disease in one case and enterectomy in the other.[5,6]

Simultaneous bilateral femoral neck fracture is a rare injury. It can occur especially when there are accompanying metabolic bone diseases such as osteomalacia, primary hyperparathyroidism and renal osteodystrophy.[7] Also, pregnancy, corticosteroid and antacid drugs and pelvic irradiation are other rare causes.[1,7]

The majority of the cases in the literature were treated with internal fixation or hemi-arthroplasty; however, some cases were followed conservatively.[8,9] Although complete recovery was achieved in some of these cases with conservative treatment, arthroplasty had to be done for the other cases due to nonunion and displacement.[8,10] As the morbidity of the patient increases with nonunion, we treated our patients with internal fixation similar to many of the cases defined in the literature.[1,5]

Avascular necrosis, refracture, delayed union and nonunion are some of the complications associated with displacement and delayed diagnosis. The main determinants of outcome of femoral neck fracture are displacement ratio and early surgical treatment.[1] The diagnosis was delayed; however, there was no displacement. Therefore, union was achieved in all patients after surgery and no avascular necrosis was seen in any of the patients.

In patients with bone pain, non traumatic fractures and muscle weakness, serum vitamin D, ALP levels and BMD should be measured for evaluation of osteomalacia regardless of age. MRI or bone scan of bilateral hips should be done in order to evaluate underlying fatigue fracture in patients with groin pain and difficulty walking even when plain X-ray is normal. The diagnosis and treatment of underlying etiology of osteomalacia is very important to improve the prognosis. In the light of these three cases, we conclude that celiac disease should be evaluated as the underlying cause of osteomalacia, especially in patients with accompanying iron deficiency anemia.

Conflict of interest

The authors declare that they have no conflicts of interest concerning this manuscript.

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