

Femoral Neck Fatigue Fracture as the First Manifestation of Celiac Disease: A Case Report

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Young individuals with fatigue fractures are often a result of the repetitive athletic activity. Military recruits, distance runners, and dancers are commonly affected and are at an increased risk of developing stress fractures in the hip. We report a case of fatigue fracture of the femoral neck in a 20-year-old software professional as the first presentation of celiac disease. The skeletal manifestation of celiac disease is rare (0.6%). The case was managed with *in situ* fixation with cannulated screws and gluten free diet. The patient was pain free with no evidence of osteonecrosis, nonunion, or any other complication at 1-year follow-up.

Keywords: Celiac disease, Fatigue fracture, Osteomalacia, Osteoporosis

INTRODUCTION

The fatigue fracture of the femoral neck in a young female with a sedentary job is a rare condition. Celiac disease is a fairly common autoimmune disorder with primarily gastrointestinal manifestations. It causes intestinal atrophy leading to malabsorption and its clinical presentations. Extra-intestinal manifestations are rare such as iron deficiency anemia, bone pains, metabolic bone diseases, and fragility fractures. Celiac disease causes osteomalacia due to decreased absorption of calcium and Vitamin D.¹ We report a rare case of fatigue fracture of the femoral neck in young female as the first presentation of celiac disease.

CASE REPORT

A 20-year-old female, a software professional, presented with groin pain in the right side for 2 months. Pain initially was mild in intensity, dull aching, but over the period, it increased to a level that patient has to walk with a limp. There was no history of trauma. A standard

anteroposterior and lateral radiographs were done which revealed break in the cortex on the superior aspect of the neck of the femur. The patient was admitted and further investigated for the fatigue fracture of the neck femur. The computed tomography scan and magnetic resonance imaging scan were done to rule out fatigue fracture on the opposite side (Figures 1 and 2). Hematological investigations revealed anemia (hemoglobin - 7.3 g%) and very low Vitamin D3 level (<3.00 ng/mL; range 30.0-100.0). Additional investigations were asked for and it revealed decreased serum calcium level of 7.5 mg/dL (range 8.8-10.6 mg/dL) with normal serum phosphorus 3.3 mg/dL (range 2.5-4.5 mg/dL). The alkaline phosphatase was markedly high 397 (range 30-120 U/L). Serum parathyroid hormone was increased to 99.8 pg/mL (range 14-72 pg/mL). All the investigations led to the diagnosis of osteomalacia. Due to deficiency of both iron and Vitamin D possibility of celiac disease was kept and tissue transglutaminase immunoglobulin A was found to be >300 (>15 U/mL suggestive of celiac disease). Biopsy was taken from duodenum for confirmation of celiac disease which showed diminished duodenal folds and histopathological confirmation was done for celiac disease.

Management

The patient was operated under regional anesthesia with fixation using two cannulated screw (DePuYSynthes) (Figure 3). The patient was advised partial weight bearing for 3 months with crutches followed by full weight bearing

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www.ijsscr.com

Month of Submission : 01-2016
 Month of Peer Review : 02-2016
 Month of Acceptance : 03-2016
 Month of Publishing : 03-2016

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Figure 1: Arrow showing fatigue fracture

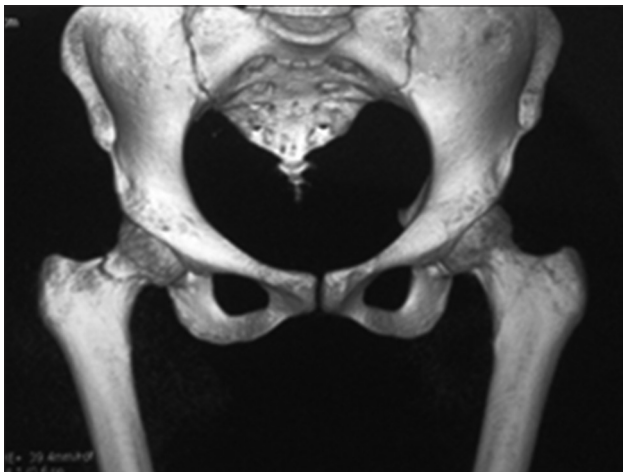


Figure 2: Three-dimensional reconstruct



Figure 3: At 1-year fracture united

with a stick. The patient was advised gluten free diet and regular follow-up was done. Her hematological values improved over the period of 3-month. At 1-year follow-up,

her fracture has united, her hemoglobin was 13.4 g%, Vitamin D level was 75.7 ng/dL, and she was pain free.

DISCUSSION

Young individuals with fatigue fractures are often a result of the repetitive athletic activity. Military recruits, distance runners, and dancers are commonly affected and are at an increased risk of developing stress fractures in the hip. The fatigue fractures occur in the elderly population with osteomalacia, osteoporosis, diabetes, rheumatoid arthritis, hyperparathyroidism, or radiation.²

When normal stress applied to abnormal bone, an insufficiency fracture occurs. Elderly women with osteoporosis tend to develop these types of injuries; however, any disorder that results in diminished resistance to physical forces as a result of defective bone health may result in an insufficiency fracture. Conversely, the normal bone that is subjected to excessive stress can develop insufficiency fractures. The repetitive submaximal stress causes bone remodeling. With increased stress, remodeling of the bone does not occur sufficiently to accommodate for the rapid breakdown of the bone that can eventually lead to stress fracture.³

Celiac disease affects bone mineral metabolism by a number of mechanisms. The classic forms of celiac disease with evident malabsorption cause decreased in plasma calcium and Vitamin D levels. These deficits lead to secondary hyperparathyroidism, which in turn increases the remodeling of the bone that results in decreased bone mass, changes the quality of bone, with the consequent decrease in strength of bone, and increases the risk of fractures. The atypical form which is mild, asymptomatic form decreases the bone mass by a different mechanism related to the secretion of inflammatory cytokines. The inflammatory cytokines increase the bone resorption by acting directly on the osteoclasts.⁴

The treatment option in fatigue fracture of the neck of femur in literature varies from conservative to internal fixation and hemiarthroplasty depending on the age, duration, and displacement of the fracture fragment.^{5,6}

Our patient presented 2 months after the onset of the pain, after investigation a diagnosis of fatigue fracture of neck of the femur was made secondary to metabolic bone disease caused by celiac disease, which was diagnosed during her stay in the hospital. There was no family history of the disease. After initiation of gluten-free diet, the patient showed remarkable improvement both in her hematological and clinical profile. At 1-year follow-up, her fracture has

healed with no evidence of osteonecrosis or any other complication.

CONCLUSION

In a young patient with fatigue fracture, bone pain, muscle weakness, decreased serum Vitamin D3, increased alkaline phosphatase, increased parathyroid hormone, and decreased bone mineral density along with iron deficiency anemia, celiac disease should be kept as possibility.

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How to cite this article: Thakur A, Sood R, Gupta A, Thakker V, Thakur R. Femoral Neck Fatigue Fracture as the First Manifestation of Celiac Disease: A Case Report. *IJSS Case Reports & Reviews* 2016;2(10):13-15.

Source of Support: Nil, **Conflict of Interest:** None declared.