The study reports a case of 14-year-old female child who was not participating in any athletic activities, and presented with gradual onset of left side hip and groin pain since 8 months duration. Clinical and radiological evaluation showed medial-sided femur neck insufficiency fracture associated with vitamin D deficiency. Femur neck insufficiency fractures are rare entity in children; usually result from overuse injuries in athletes. Absence of athletic activities raises the suspicion of endocrine or metabolic derangement as an underlying cause of insufficiency fracture.

Key words: Femur neck, insufficiency fracture, adolescent, vitamin D deficiency.

INTRODUCTION

Stress fractures represent the inability of the bone to withstand the repeated load exerted from athletic activities or even normal daily life events on a normal or weakened bone, respectively. This repetitive load results in microdamage to the bone, which may progress and result in fracture; if the healing process does not proceed properly (Kaeding and Miller, 2013).

Stress fractures have different severity of structural changes and different potential for healing related to their location and etiology. Femur neck stress fractures are extremely rare in children with open proximal femoral physis, but should be included in the differential diagnosis of hip pain in children, in addition to synovitis, infection, slipped capital femoral epiphysis, Leg-Calves-Perthes disease and benign and malignant neoplasms (Er et al., 2014). All cases of femur neck stress fractures published in the literature were either in athletes or in children participating in repetitive high impact activities (Er et al., 2014; Lehman and Shah, 2004). None of them highlighted a vitamin D deficiency as an associated finding of their patient’s problem. One publication focused on the metabolic pathology as an underlying cause of stress fractures in the tibia, fibula and femoral shaft (Turturro et al., 2010), but none showed an insufficiency fracture in the femur neck of a child or adolescent.

The study presents the case of a fourteen-year-old female patient with insufficiency fracture at the medial side of femur neck associated with vitamin D deficiency. To this study knowledge, this is the first report of a looser’s zone in the femur neck associated with vitamin D deficiency in an adolescent. The patient and her parents gave consent to the publication of this report.

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A fourteen-year-old female patient was presented to Pediatric Orthopedic Clinic with symptoms of left groin pain and limping since 8 months. The pain started gradually and become progressive with time. The patient denied any recent trauma or participation in any vigorous or sport activity prior to her complaint. She had her menarche at the age of thirteen with regular cycles. She does not get enough sun exposure during the daytime, which is related to cultural reasons, nor get enough vitamin D or calcium-containing dairy products or supply. Her past medical history was remarkable for tonsillectomy and adenoidectomy at the age of 2 years.

Her vital signs were normal including a temperature of 37.2°C and a pulse rate of 78. On physical examination, there was painful left hip motion and a negative test for obligatory external rotation when the left hip is passively flexed to 90°. The left leg had normal muscle strength, with no signs of infection. Laboratory investigations, including complete blood cell count and kidney and liver function tests were normal. C-reactive protein level was less than 1 mg/L (normal, less than 5 mg/L), and normal erythrocyte sedimentation rate of 20 mm/h (normal, 0 to 20 mm/h).

Hips and pelvis radiography showed cortical defect surrounded with sclerosis at the medial side of the left femoral neck, but normal femoral head contour noted with no signs of avascular necrosis or slipped capital femoral epiphysis.

However, no nidus was apparently clearly seen on radiography, but osteoid osteoma was included in the differential diagnosis (Figure 1). Accordingly, hips computed tomography (CT) scan (Figure 2) was performed, and revealed cortical disruption at the medial aspect of left femoral neck surrounded with minimal sclerosis, which raised the possibility of insufficiency fracture. Magnetic resonance imaging (MRI) of the hips and pelvis was performed and T1WI showed linear hypo-intensity (Figure 3), while T2WI including fat saturation showed linear hypo-intensity surrounded with high signal intensity (Figure 4) due to bone marrow and soft tissue edema at the medial side of the left femoral neck. These findings confirmed the diagnosis of insufficiency fracture (looser's zone).

The clinical history did not show any risk factor of overuse or change in the patient life style. Therefore, metabolic work up was carried out to rule out pathological changes that can lead to stress fractures (looser's zone). Accordingly, chemical and endocrine tests were performed and revealed decreased serum calcium level of 7.2 mg/dl (normal 8.5 to 10.5 mg/dl), decreased serum phosphorus level of 2.7 mg/dl (normal 2.7 to 4.5 mg/dl), decreased 25-hydroxyvitamin D (25OHD) level of less than 3.5 ng/ml (sufficiency: 30 to150 ng/ml), high serum level of parathyroid hormone of 339.9 pg/ml (normal 9 to 55 pg/ml), high serum alkaline phosphatase level of 311 U/L (normal 30 to 120 U/L), normal magnesium serum level of 2.4 mg/dl (normal 1.58 to 2.55 mg/dl), normal thyroid function test, normal follicle-stimulating hormone level, normal 17-beta-estradiol and normal serum cortisol level.

The patient was diagnosed as having vitamin D deficiency with secondary hyperparathyroidism, which might induce femur neck insufficiency fracture and she was started on Vitamin D treatment (50 000 IU once weekly for 8 weeks and then 5000 IU every other day for
Figure 3. Coronal MRI T1W image showing the stress fracture as linear hypo-intensity (arrow) at the medial side of left femoral neck.

Figure 4. Coronal MRI T2W image with fat saturation showing the stress fracture as linear hypo-intensity (arrow) surrounded with bone marrow and soft tissue edema seen as signal high intensity (arrow head) at the medial side of the left femoral neck.

8 weeks) and then she was maintained on 50 000 IU once monthly), and calcium therapy 500 mg PO q8 hours, in addition to non-weight bearing using crutches and pain killer medications. 18 months later the patient walks with full weight bearing without pain, her laboratory investigations showed normalized calcium level of 9.3 mg/dl, normal phosphorus level of 4.1 mg/dl, normal alkaline phosphatase level of 79U/L, improvement of her 25OHD level to 34 ng/ml and parathyroid level has decreased to 128.2 pg/ml.

Hips and pelvis radiography showed healed cortical defect with no apparent cortical disruption at the medial side of the femoral neck (Figure 5). Magnetic resonance imaging (MRI) of the hips and pelvis was performed and T1WI showed almost complete resolution the linear hypo-intensity (insufficiency fracture) at the left femoral neck (Figure 6), while T2WI (Figure 7) including fat saturation showed absence of the linear hypo-intensity which was seen in Figure 4, with disappearance of the high signal intensity due to bone marrow and soft tissue edema at the medial side of the left femoral neck.

DISCUSSION

The first femoral neck stress fracture was reported by Devas in an under nourished 14-year-old boy who also
had other stress fractures in the tibia and unilateral avulsion of the ischial tuberosity. He described two types of femoral neck stress fractures (Devas, 1961). First, the transverse type, which is located in the superior tensile part of the femoral neck, and has the tendency of progression, that makes its operative fixation mandatory. The second is the compression type, which is located in the inferior aspect of the femoral neck that has the tendency to heal non-operatively, if the stressful load is relieved (Wolfgang, 1977).

Common disorders that present with hip or groin pain are infection, synovitis, slipped capital femoral epiphysis, leg-calves-perthe’s disease, trauma and tumors (Er et al., 2014). In the pubertal stage of development, a common cause of hip pain is a slipped capital femoral epiphysis which was ruled out in our patient due to the normal alignment of the femoral head and neck, and the absent of peri-physeal edematous changes as seen on the MRI of the proximal femur. The other causes were ruled out based on the normality of initial laboratory blood results and the radiological evaluation.

On the basis of absence of typical presentation of femur neck stress fracture with repetitive load, and the absence of signs of healing after 8 months of complaints, the study believed that an underlying endocrinologic or metabolic cause should be present to interpret the present situation. Er et al. (2014) in their literature review and Fiévez et al. (2013) stated that “there were no reports of any underlying medical conditions for a femoral neck stress fracture in children”. To the contrary, this study found through the work up which was done for the study patient a metabolic derangement manifested by a very low level of serum 25OHD (less than 3.5 ng/ml), low serum level of both calcium and phosphorus, high serum alkaline phosphatase level with secondary increase in the parathyroid hormone, representing a well-established case of vitamin D deficiency.

Conclusion
Femur neck stress fractures are a rare entity in immature children (Kaeding and Miller, 2013). There are less than 20 cases reported in the literature, with female to male ratio of 2.2 to 1, and both sides are affected equally (Table 1). Most of them showed high athletic activities as a risk factor for the fracture development. None of them came through any endocrinologic or metabolic abnormalities that may be associated or precipitate to a femoral neck stress or insufficiency fracture. An accurate diagnosis depends on a systematic approach starting from the clinical history, physical examination and then proper investigations guided by the clues from the patient’s clinical evaluation. To the best of this study knowledge, this is the first report of a looser’s zone in the femur neck in an adolescent associated with Vitamin D deficiency (Osteomalacia).

Conflict of Interests
The authors have not declared any conflict of interests.
Table 1. Literature review (20 cases) of femur neck insufficiency fracture in a skeletally immature child.

<table>
<thead>
<tr>
<th>Study</th>
<th>Age (Y)</th>
<th>Sex</th>
<th>Side</th>
<th>Type</th>
<th>Underlying metabolic cause</th>
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<tr>
<td>Wolfgang, 1977</td>
<td>10</td>
<td>F</td>
<td>Right</td>
<td>Compression</td>
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<td>Miller, 1979</td>
<td>14</td>
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<td>Left</td>
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<td>F</td>
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</tr>
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<td>Scheerlinck and DeBoeck, 1998</td>
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<td>8</td>
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<td>M</td>
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</tr>
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<tr>
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<td></td>
<td></td>
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REFERENCES


