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Case Report

Uncommon presentation of bilateral subtrochanteric insufficiency fractures in young male associated with hypogonadism: A case report

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ABSTRACT

Hypogonadism in men is a well-recognized cause of secondary osteoporosis. It is characterized by insufficient production of androgen, testosterone, and sperms. Testosterone deficiency is the key factor for insufficiency fractures in men, resulting from normal loading on an osteoporotic bone. In this report, we are presenting a case of a 27-year-old male known to have hypogonadotropic hypogonadism, who suffered from both femoral atraumatic subtrochanteric fractures that had been existing for a considerable time. The fractures interfered with the patient's walking and daily activity. It is worth reporting this case because most of the proximal femur insufficiency fractures mentioned in the literature correlated with prolonged alendronate therapy or were post-bariatric surgery.

Keywords: Femur, Hypogonadism, Insufficiency fractures, Stress fracture, Subtrochanteric

INTRODUCTION

Hypogonadism is characterized by insufficient production of androgen and testosterone; it is a well-recognized cause of secondary osteoporosis in men affecting the microarchitecture of the bone cortices and trabeculae that increase the tendency to fragility fractures reflected by decreased bone mineral density (BMD). The severity varies depending on the duration of the disease.[1] Androgen and testosterone positively influence the bone size, mass, and remodeling.[2] Androgen exerts a direct influence on bone formation, resorption, and homeostasis through its receptor on bone-related cells in addition to progenitor cells.[2,3] Its key role is in helping the proliferation and differentiation of osteoblasts.[2] On the other hand, testosterone directly impacts bone health, promoting bone size and mass by stimulating osteoblasts to produce trabecular bone and osteoprotegerin, decreasing the osteoclast formation, and motivating the osteocytes to prevent trabecular bone loss.[1-3]

Furthermore, the effect of testosterone extends to increase chondrocytes development, metaphyseal ossification, periosteal new bone formation, and long bone growth by raising the

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calcium retention and incorporation into the bone.[1-3] Unfortunately, the specific testosterone role in bone health maintenance and the development of osteoporosis in men when deficient is not completely clear.[1] Moreover, testosterone deficiency does not predict fracture risk.[4] Androgen and testosterone deficiency manifests as osteopenia or osteoporosis. It is related to the reversible reduction in BMD that can be statistically and significantly corrected with testosterone therapy over a short period and minimizing the risk of fractures in men.[1,5]

Several overlapping terminologies have been used to explain non-traumatic fractures, including stress, insufficiency, fatigue, and fragility, which hindered understanding and reporting these injuries.[6] In the term's broadest meaning, stress fractures include fatigue and insufficiency fractures. A fatigue fracture is a localized failure in the normal bone due to repetitive applied stress, usually occurring with an abruptly increased intensity, duration, or frequency of activity when osteoclasts activity is greater than osteoblasts among young, athletic people and in females more than males.[6-8] On the other hand, insufficiency and fragility fractures are secondary to normal stress on the abnormally weakened bone (osteomalacia).

Insufficiency fractures are frequent among many medical conditions, with osteoporosis being the common entity between them. Some of the medical conditions are metabolic bone disease, rheumatoid arthritis, neurological disorders, total hip replacement, corticosteroid therapy, previous irradiation, and high-dose fluoride therapy.[7]

Osteoporosis is diagnosed when a BMD lies 2.5 standard deviations (SD) measurements or less than the average value for young adult Caucasian healthy women.[1,9] Accordingly, using a female reference underestimates male bone loss and extends the diagnosis of osteoporosis.[10] Males and females have the same fracture risk for a given BMD value.[1,11] Therefore, regardless of utilizing a female or male reference, BMD T-score SD is used to define osteoporosis and osteopenia in males aged equal to or above 50 years. For individuals under 50 years, the BMD Z-score is more applicable.[1] Consequently, greater appreciation and awareness of the importance of male hypogonadal osteoporosis enables us to understand men's bone quality.[9]

Dual-energy X-ray absorptiometry (DEXA) is a first choice non-invasive, accurate, and cost-effective test. In addition, the diagnostic laboratory workup such as serum calcium, phosphorus, alkaline phosphatase, 25-hydroxyvitamin D levels, C-terminal telopeptide, renal function tests, liver function tests, and 24 h urine calcium is usually used for evaluating bone density in men.[1]

The imaging features of stress fractures are defined according to the relative proportion of trabecular to cortical bone and the injury chronicity. The initial radiographic sign in the cortical bone is a subtle cortical lucency at the site of the microfracture and the osteoclast resorption followed by cortical thickening, endosteal callus production, and periosteal reaction; the injury propagates until the cortical breaking line appears evident.[6,12] In the trabecular bone, the earliest sign is subtle faint and blurring sclerosis progresses to linear intramedullary sclerosis, representing a microcallus production and a new bone deposition along with the trabecular remodeling.[6,12]

The present report aims to present a young man diagnosed with hypogonadotropic hypogonadism who suffered from bilateral subtrochanteric insufficiency fracture. Up to our knowledge, this is considered an unusual cause for the fracture; as per the literature, most femur insufficiency fractures are caused by prolonged use of alendronate.

CASE REPORT

We are presenting the case of a 27-year-old male working as a teacher. He is a known case of hypogonadism who has been followed by an endocrinologist on regular medications (chorionic gonadotropin injection, clomiphene, and anastrozole). Otherwise, he was medically free and was not on alendronates therapy. The patient presented to the orthopedic clinic suffering from a long-standing pain in the inguinal region and upper thigh for more than 10 years without noticeable trauma. Late presentation to the orthopedic clinic could be due to the slow course of the disease and the initiation of the medical treatment.

He described the pain as dull and bony, more frequent during the early morning, walking, and standing for a long time. The pain was preventing him from his daily activities. He claimed that his symptoms were relieved by rest and paracetamol. Although from the start of his complaint before 10 years, he was evaluated by several physicians who treated him symptomatically with analgesia and supplements; the last physician referred him to the endocrine clinic 7 years back as a case of delayed puberty and infertility. His blood workup results showed a low testosterone level. As a result, he was commenced on testosterone therapy. In addition, growth hormone treatment was started as his radiograph indicated delayed physeal plates closure. On examination, he had short stature, with bilateral tenderness on the proximal femur and groin regions. In addition, he had a global limitation of hip joints motion, mainly in flexion and abduction. Furthermore, he had a painful motion on the right side more associated with weak hips abductors making him walk with a lurching gait. No further positive findings. His lower extremities' sensory, motor, and vascular examinations were normal. The plain radiographs revealed decreased cortical thickness and loss of bony trabeculae, indicating decreased BMD. Furthermore, there were transverse cortical lucencies in the

subtrochanteric regions, complete non-displaced on the right femur, and incomplete at the lateral cortex of the left femoral shaft, demonstrating an insufficiency fracture in these areas [Figure 1].

There was an improvement in the testosterone level, but it did not reach the normal range. His prolactin level was reduced but still was above the normal value. On the other hand, follicle-stimulating hormone and luteinizing hormone were markedly elevated above the normal value. Calcium levels did not improve despite the quantitative improvement in Vitamin D. The thyroid profile tests were normal.

Unfortunately, a DEXA scan was not done during his treatment with the endocrinologist but done after his first visit to our clinic. Nevertheless, his BMD demonstrated marked osteoporosis, as the DEXA scan was –3, –3.3, and –3.8 T-scores of the lumbar spine, right, and left proximal femur, respectively, while the Z-scores were –3, –3.3, and –3.7.

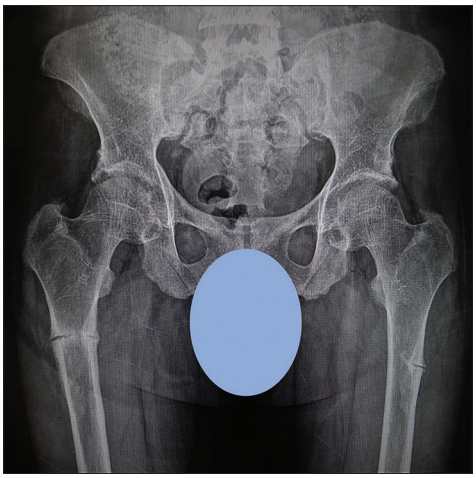

Figure 1: The anterior-posterior radiograph of the pelvis, both hips, and proximal femora demonstrates a complete non-displaced subtrochanteric right femur fracture and an incomplete non-displaced fracture of the left side. Blue oval shape was added to cover the private region.

Figure 2: Post-operative anterior-posterior radiograph of the right hip and proximal femur demonstrates trochanteric intramedullary femoral nail.

The decision was that the patient needed prophylactic intramedullary femur nailing followed by vigorous physical therapy to strengthen the hip abductors and regain the range of motion. Accordingly, the patient was admitted, and a consultation of the internal medicine and endocrine specialties was made to correct his electrolytes abnormalities and optimize his medical status. Subsequently, the patient was taken to the operating room for trochanteric entry femur nailing fixation for the right femur. We operated on the right side first because it is a complete fracture and more symptomatic regarding the pain. The procedure was performed under spinal anesthesia and on a traction table through standard technique [Figure 2]. He was advised to walk with weight-bearing as tolerated. Dietician service was consulted, who followed up the patient during his admission. At his first post-operative follow-up visit at 2 weeks, he was doing great, and his sutures were removed. The patient was referred to physical therapy. Before submitting the present report, his hips abductors were stronger, increasing both hips abduction and flexion associated with pain and lurching gait improvement, and the last post-operative radiograph was done in the last visit 6 months after surgery [Figure 3].

DISCUSSION

Pain with activity relieved by rest is the main presentation of insufficient fractures. Its severity changes according to the activity level. Lower limbs are the most common site of the complaint and it is uncommon in the upper extremity.[6,12] In our case, the patient complained of pain in his hip when walking, with his medical condition raising the suspicion of insufficient fractures, which were confirmed by the radiological examinations. Moreover, the presentation

Figure 3: Anterior-posterior radiograph of the right hip and proximal femur 6 months after surgery.

of bilateral subtrochanteric insufficiency fractures in young male due to hypogonadotropic hypogonadism, which is unprecedented considering that the fractures were caused by daily activities. Although we do not have a clear explanation for the unique finding, simultaneous bilateral subtrochanteric fracture. In addition, with the duration of his symptoms, he continued weight-bearing on both lower limbs. The medial and lateral sides of the femur have stresses that resulted in a complete fracture through both cortices, transverse, and non-comminuted, consistent with insufficiency fractures. Although the plain radiograph is the first line for musculoskeletal pathologies imaging, its sensitivity to detect early-stage insufficiency fractures is about 15–35% and 30–70% for the late stages.[6,7] Instead, magnetic resonance imaging has a 100% sensitivity and an 85% specificity, which is considered a second-line modality, requested when the radiographs are normal, or the pain etiology is unknown.[6,13,14]

Computed tomography is useful for detecting longitudinal fracture lines and stress fractures of the spine. However, recently, it is not considered to be an imaging modality for the diagnostic evaluation of suspected stress fractures.[6,7] Technetium-99m-methylene diphosphonate (Tc-99m-MDP) bone scans are often considered the gold standard modality due to its sensitivity in detecting metabolic bone abnormal activity, but it has a low specificity rate.[7,13]

Osteoporosis is the most common underlying etiology causing insufficiency fractures. Many disorders could develop insufficiency fractures that influence the bone's ability to withstand normal loading forces. However, most of the cases in the literature indicate the association with the long-term use of bisphosphonate.

Early recognition, prevention, and treatment should be initiated by realizing the burden of osteoporosis›s consequences. The ideal therapeutic strategies should start with identifying patients at risk and initiating preventative measures before the occurrence of insufficiency fractures. The preventive measures are multidisciplinary approaches to initiate behavioral modification, education, and exercises to increase awareness of the bone mineral disorder and its risk. DEXA scan is the cornerstone in the early detection of bone mineral abnormality. Medical therapy should be commenced immediately with hormone replacement therapy or anti-resorption drugs accordingly when indicated.

The treatment of the insufficiency fractures once occurred depends on the fracture location, extent and displacement, the functional status, and the coexisting other medical diseases.

The endocrinology society male osteoporosis guidelines, 2012, recommended a combination therapy with a bisphosphonate or teriparatide with the testosterone replacement in high-risk cases to enhance BMD and reduce the fracture risk for all 50 years men and older patients with a hip or spine fragility fracture, osteopenia, or osteoporosis.[15] Unfortunately, the diagnosis and the treatment indications remain poorly defined for osteoporosis in young adults between 20 and 50 years of age.[16] Although bisphosphonates and denosumab are helpful in raising BMD in hypogonadal men but are more frequently utilized in older men.[16]

Testosterone therapy in hypogonadal males remarkably improves in BMD, mainly in arms, hip, and femur, except for the lumbar spine.[1,15,17] Despite that, no studies have evaluated its clinical influence on fracture risk.[1,15,17] Studies indicated that bisphosphonate therapy positively affects femoral and lumbar BMD associated with a significant reduction in fractures.[1] Despite that, the endocrinology society has encouraged using denosumab and teriparatide as an alternative to bisphosphonate therapy in males with treatment indications. Their efficacy in the hypogonadal male population is yet to be studied.[1,15] Intramedullary nailing is the preferred treatment when surgery is indicated and could be considered as the standard of care for all oblique or transverse diaphyseal fractures of the lower extremity.[6]

CONCLUSION

The insufficient fracture should be considered and ruled out when the hypogonadotropic hypogonadism patient complains of walking hip pain when walking, and a plain radiological examination must be requested. A walker patient with full weight-bearing is not an exception to the presence of a complete subtrochanteric fracture through both cortices.

AUTHORS' CONTRIBUTIONS

WR, EA, and AS conceived and designed the study and wrote the initial draft of the article. RA and YA collected, organized, analyzed, and interpreted the data. SS provided the critical revision and wrote the final draft of the article. All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

DECLARATION OF PATIENT CONSENT

The authors' certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initials will not be published and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

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CONFLICTS OF INTEREST

There are no conflicts of interest.

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