Olgu Sunumu / Case Report
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A case of stress fracture: What is the underlying cause?

Bir stres kırığı olgusu: Altta yatan neden nedir?

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ÖZET

Stres kırıklarına klinik pratikte sıkça rastlanılmaktadır. Stres kırıklarının birçok özelliği ortaktır, ancak altta yatan kemiğin durumuna bağlı farklılıklar vardır. Klinisyen stres kırığı tipine göre yaklaşımda bulunmalıdır. Bu makalede uzun süreli bifosfonat tedavisi ve tek doz denosumab alan, sekiz ay arayla bilateral atipik femoral kırığı gelişmiş 71 yaşında osteoporozlu bir kadın hastayı sunuyoruz. Doğru tanı koymak için klinik değerlendirmenin radyografik, dansitometrik ve biyokimyasal bulgularla birleştirilmesi gerekir.

Anahtar Kelimeler: stres fraktürü, atipik femur fraktürü, bifosfonat, denosumab

ABSTRACT

Stress fractures are encountered commonly in clinical practice. Many features of stress fractures are in common, but the differences depend on the condition of the underlying bone. The clinician should approach according to the type of stress fracture. We report a 71-year-old female patient with osteoporosis, who had been taking prolonged bisphosphonate therapy and a single dose denosumab, sustained bilateral atypical femoral fractures (AFF) eight months apart. Collating the clinical assessment with the radiographic, densitometric and biochemical findings is needed in making accurate diagnosis..

Keywords: stress fracture, atypical femoral fracture, bisphosphonate, denosumab

INTRODUCTION

Stress fractures are repetitive strain injuries. Many features of stress fractures are in common, but the differences depend on the condition of the underlying bone. There are two types, fatigue-type in normal bone and the insufficiency-type in abnormal bone. Here, we aimed to present an elderly female patient with osteoporosis, who had been taking prolonged bisphosphonate therapy and a single dose denosumab, sustained bilateral insufficiency-type stress fractures eight months apart..

CASE

A 71-year-old female patient with 8-year history of post-menopausal osteoporosis presented with right thigh pain and waddling gait. She took prolonged oral bisphosphonate therapy (alendronate for six years and then oral ibandronate for one 1 year) without drug holiday. She had been admitted in September 2018 with a low-trauma femur fracture on the opposite (left) side, and recalled that she had complained of mild left thigh pain at the time (Figure 1). Bone mineral densitometry of the spine and bilateral hips was measured in January 2019. Because the T score was -2.9 at the spine L1-L4 and -3,4 at the right hip, oral ibandronate therapy was stopped and subcutaneous injection of denosumab was started. Calcium and vitamin D supplementation continued.

Physical examination revealed a painful movements of lower extremities with intact neurovascular exam. Laboratory and radiological studies were performed. Her serum-corrected calcium, magnesium, phosphate, alkaline phosphatase levels are in normal ranges, but paratyroid hormon level was 364 pg/mL, 25-OH vitamin D 5,6 ng/mL. Radiographs revealed two focal thickenings of the lateral diaphyseal cortex on the right femur and a incomplet femoral shaft fracture with lateral cortical (periosteal and endosteal) hypertrophy and beaking on the left femur (Figure 2).

Initially, denosumab was stopped. The patient was consulted with orthopedic clinic for fracture and endocrinology clinic for elevated parathyroid hormone. Vitamin D supplementation was recommended by the endocrinologist. Parathormone tended to decrease and vitamin D to increase in control blood tests. After 3 months

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of treatment, her parathormon level was 101 pg/mL, 25-OH vitamin D 47,7 ng/mL. The patient refused to have surgery. Limited activity with partial weight-bearing was recommended. The patient was informed about possible symptoms and followed up regularly. At the time of writing, she was awaiting endocrinologic assessment for teriparatide therapy.

Figure 1.*Left femur. Focal lateral cortical thickening on the radiograph (A) and periosteal high signal on the T2-weighted MRI (B) .*

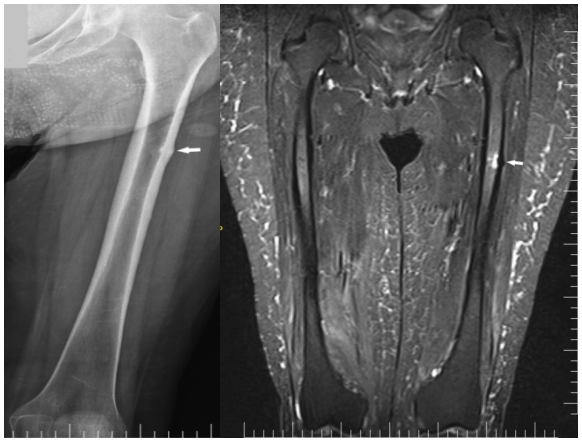
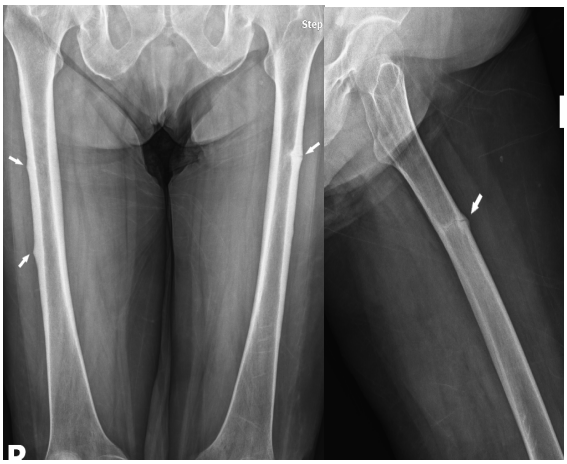


Figure 2. *Radiographs showed 3 focal thickenings (A) of the lateral diaphyseal cortex, 1 of them with a visible crack (radiolucent line >1/2 of bone diameter) on the left femur (B).*



DISCUSSION

Stress fractures are encountered commonly in clinical practice and lie along a spectrum. In some cases, it can be difficult to differentiate between them. The fatigue-type usually relates to activity level in young adults without bone disease. Insufficiency-type stress fracture might be of many different types; looser zones of osteomalacia, atypical femoral fractures (AFF) in osteoporosis, Paget's disease of bone, rare disorders like hypophosphatasia. Stress fractures appear on the convex surface of the shaft of long bones due to tensile stress in AFF and paget's disease , while on the concave surface due to compressive stress in osteoporosis and osteomalacia (1).

Looser zones of osteomalacia usually occur multiple, symmetric and on the medial side of the femur. Typically, they appear a broad band of lucency, parallel margins, marginal sclerosis, callus (if on treatment) and delayed healing (2). Continuously elevated parathyroid hormone has little impact on the trabecular bone and a decrease in the cortical bone. Thickening of the lateral cortex were not consistent with bone changes in hyperparathyroidism (3).

Atypical femoral fractures are located along the femoral diaphysis from lesser trochanter to supracondylar flare.They can be recognised by the presence of major and minor features, described in 2010. AFF is diagnosed based on subtrochanteric or femoral shaft location and the presence of at least four the following five major features: minimal or no trauma, fracture originating at the lateral cortex and being substantially transverse, complete fractures extending through both cortices, localised periosteal or endosteal cortical thickening and minimal or no comminution. Minor criterias; cortical thickness of the femoral diaphysis, bilaterality, prodromal symptoms and delayed healing aren't necessary for diagnosis, but they has associated with fracture (2). Our case met all of major and minor criterias. Osteoporosis, hyperparathyroidism and vitamin D deficiency may have increased susceptibility to fractures, but the main cause is bisphosphonate and denosumab exposure in our case.

The incidence of AFFs varies depending on the duration of bisphosphonate use (2 / 100,000 cases per year after 2 years of bisphosphonate use - 78 / 100,000 cases per year after 8 years of use) (4). The patient should be reassessed at 3 to 5 years to determine if a drug holiday should be initiated. Prodromal pain,even after treatment has stopped, is a warning symptom of impending fracture. If it occurs, imaging studies should be done (5).It is not known exactly which way bisphosphonates cause AFFs. Over-suppresssion of bone turnover, accumulation of microcracks in the bone over time, fragility due to increased bone mineralization are the mechanisms accused in etiopathogenesis. The relationship between bisphosphonate treatment and non-femoral fractures is still unclear (1).

Approximately 25% of AFF cases are predicted to develop fractures in the opposite femur. Controlateral fracture occurs from 1 month to 4 years after the index AFF (6). Based on a observational study, the risk of sustaining a contralateral fracture if bisphosphonate exposure is stopped soon after the index fracture is in the range of 18 percent. If bisphosphonate therapy is continued, the risk of contralateral fracture rises over time, to 25.8 percent at 1 year and to 53.8 percent at 3 years after the index atypical femur fracture (7). Min et all. have developed a scoring system to assess the risk of progressing incomplet fractures to complet fractures and recomended the follow-up of patients scoring 8 or less and the prophylactic intramedullary nail fixation of others (8). The first score of our case was 8 and the next was 14.

In conclusion, delay in diagnosis is a common clinical occurrence due to the insidious clinical presentation and unaware of the clinicians of the probability. Collating the clinical assessment with the radiographic, densitometric and biochemical findings is needed in making accurate diagnosis. Radiographic features of the fracture are the main aspect of the differential diagnosis.

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REFERENCES

1. McKenna MJ, Heffernan E, Hurson C, McKiernan FE. Clinician approach to diagnosis of stress fractures including bisphosphonate-associated fractures. QJM 2014 Feb; 107(2): 99-105.

2. Shane E, Burr D, Abrahamsen B, et al. Atypical subtrochanteric and diaphyseal femoral fractures: second report of a task force of the American Society for Bone and Mineral Research. J Bone Miner Res 2014; 29(1): 1-23.

3. Butler I,Tipping B, Bhaga R. Atypical femoral fractures. JEMDSA 2013; 18(2): 125-127.

4. Dell R, Greene D, Ott SM, Silverman S, Eisemon E, Funahashi T, et al. A retrospective analysis of all atypical femur fractures seen in a large California HMO from the years 2007 to 2009. J Bone Miner Res. 2010; 25: 61.

5. Dell R, Greene D. A proposal for an atypical femur fracture treatment and prevention clinical practice guideline. Osteoporos Int. 2018 Jun;29(6):1277-1283

6. Silverman S, Kupperman E, Bukata S. Bisphosphonate-related atypical femoral fracture: Managing a rare but serious complication. Cleve Clin J Med 2018 Nov; 85(11): 885-893

7. Dell RM, Greene D, Tran D. Stopping bisphosphonate treatment decreases the risk of having a second atypical femoral fracture. Paper presented at: American Academy of Orthopaedic Surgeons (AAOS) Annual Meeting 2012 Feb 7-11; San Francisco, CA, USA.

8. Min BW, Koo KH, Park YS, Oh CW, Lim SJ, Kim JW, et al. Scoring System for Identifying Impending Complete Fractures in incomplete Atypical Femoral Fractures. J Clin Endocrinol Metab 2017; 102(2): 545-50.