CASE REPORT

Atypical Fracture Shaft of Femur Case Series

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Abstract

Atypical fractures of the shaft of femur are not rare. Its incidence has been found higher in patients on long term bisphosphonate therapy. However, it also occurs in older patients who have not taken bisphosphonates. We are reporting 5 cases of atypical fracture shaft of femur, all of whom had similar history and was managed similarly with closed reduction and internal fixation with intramedullary nail. All had good union of fractures with no residual deformity.

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Introduction

Atypical femoral fractures have a distinctive radiographic appearance and can occur spontaneously with or without prodromal pain. In light of the uncertainty about the pathogenic mechanisms and controversy surrounding the association of atypical fractures with bisphosphonates, the management of patients with history of prolonged use of bisphosphonates is challenging. A comprehensive metabolic approach is necessary to prevent over suppression of bone remodeling, which has been implicated in the pathogenesis of these fractures. The American Society for Bone and Mineral Research (ASBMR) Task Force report established a provisional case definition[1] and reviewed the literature on atypical fractures.

The earliest reports raising concerns about a possible link between bisphosphonate therapy and atypical fractures described unusual low-energy subtrochanteric or femoral shaft fractures in patients with severely suppressed bone turnover.[2] However, the association between these fractures and bisphosphonate therapy was unclear because some of the patients studied were taking multiple bone-active agents, including estrogen, bisphosphonates, and glucocorticoids.[3]

We are reporting 5 cases of atypical fractures of the femur. All the patients were above the age of 65years, and had history of trivial fall with consequent pain in the thigh. Out of the 5, only 2 patients were on bisphosphonate therapy for osteoporosis; one had been on Ibandronate for one year (Figure 1), and the other on Alendronate for 3 years (Figure 3). The other patients (Figures 2, 4 and 5) did not have any history of therapy with bisphosphonates or corticosteroids. Radiological evaluation showed all the patients had atypical fracture femur (as per the description of atypical). None of them had any significant metabolic or biochemical abnormalities. Screening of the contralateral femur did not reveal any stress fractures, or impending fractures in any of these patients. All patients were managed with closed reduction and cephalomedullary nailing, with withdrawal of bisphosphonates in the two patients who were on these. Postoperatively, all were given

vitamin D and calcium. There was no delay in fracture healing in any of the patients.

Discussion

The epidemiology of atypical femoral fractures is poorly understood compared to that of typical osteoporotic hip fractures. Furthermore, because no diagnostic code currently exists for atypical fractures, studies examining only diagnostic coding cannot distinguish atypical subtrochanteric fractures from typical subtrochanteric fractures; radiographs are required to identify features of atypia.[3,4]

The association between atypical femoral fractures and bisphosphonate therapy is complex and somewhat controversial.[4] Bisphosphonate therapy appears to increase risk of atypical fractures, with greater risk associated with longer durations of treatment and declining risk after cessation of treatment. However, atypical fractures have also been observed in patients who have never been exposed to bisphosphonates. Thus, while bisphosphonate treatment may be an important risk factor for atypical fractures, it cannot be the sole risk factor.[5]

Atypical fractures appear to initiate as stress fractures on the lateral cortex of the proximal femur, a site of high tensile loads due to bending. The prodromal pain experienced by some patients with atypical fractures is consistent with an incipient stress fracture. The unusual transverse fracture morphology and lack of communition also suggest a brittle tensile failure mechanism.

At the material level, bisphosphonates alter tissue properties by reducing turnover, thereby increasing tissue age, i.e. time since tissue formation. In the organic matrix, bisphosphonate treatment allows accumulation of nonenzymatic crosslinks (advanced glycation endproducts), which is associated with reduced bone toughness, and increases the maturity of the enzymatic crosslinks.[6] Bisphosphonate-treated tissue from the lateral proximal femur had a narrower distribution of cortical collagen maturity and crystallinity compared to bisphosphonate-naïve tissue. Similarly, nano-mechanical analysis of iliac crest biopsies revealed narrower distributions of elastic modulus and hardness in cortical tissue of patients with severely suppressed bone turnover relative to controls.[3,7] Reduced heterogeneity of tissue properties may be associated with reduced toughness, although this relationship has not yet been established directly for human bone. Finally, the proximal lateral femoral cortices of patients with atypical fractures had 8% greater tissue mineral content than patients with typical fractures, consistent with the brittle failure pattern observed at this highly mineralized site.

Table 1. Diagnostic Features of Atypical Fracture

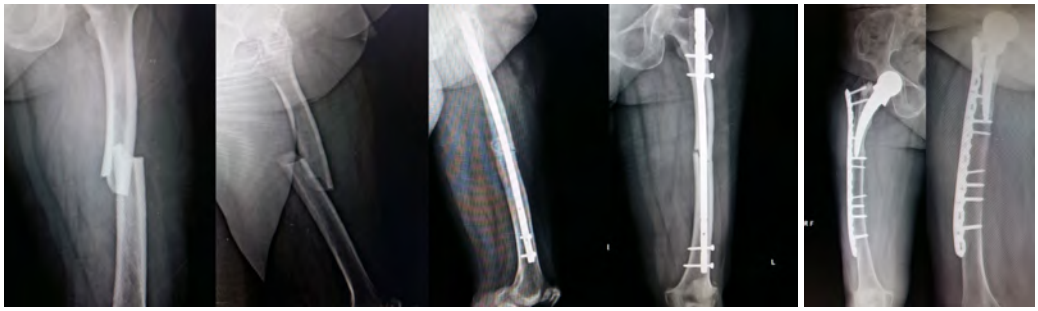
Major Features	Minor Features
No history of trauma, or associated with low-energy trauma	Localized periosteal thickening of the lateral cortex
Fracture located anywhere from distal to the lesser trochanter to proximal to the supracondylar area	Generalized thickening of the femoral cortices
Transverse or short oblique fracture configuration	Prodromal symptoms
Noncomminuted fracture	May be associated with bilateral fractures or symptoms
Medial spike in complete fractures; incomplete fractures involve only the lateral cortex	Evidence of delayed fracture-healing
	Comorbid conditions or the use of some medicationslike corticosteroids or bisphosphonates

Diagnosis

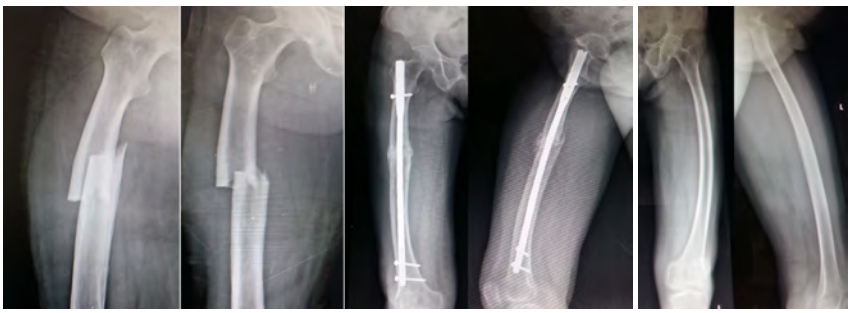
Diagnosis of an atypical fracture femur is based on radiological features. The American Society for Bone and Mineral Research (ASBMR) Task Force report established a provisional case definition and reviewed the literature on atypical fractures through 2010. All major features must be present to consider a fracture "atypical" and distinguish it from typical fractures, while minor features may or may not be present (Table 1).

Management of Patients With Atypical Femoral Fractures

Management of patients with atypical femoral fractures includes fracture fixation and initiation of medical management. An intramedullary reconstruction full-length nail is preferred because fractures treated with intramedullary nails heal by endochondral repair, which allows for stabilization via callus formation, while plate-screw constructs generally preclude the endochondral repair process.[8,9] Although minimal data are available on healing of atypical fractures, preliminary evidence suggests that healing may be impaired. Following diagnosis of atypical femoral fracture from radiographic evaluation, bisphosphonate therapy should be discontinued. Patients should receive daily calcium supplementation of 1000–1200 mg/day, and 25-hydroxyvitamin D should



Case 1



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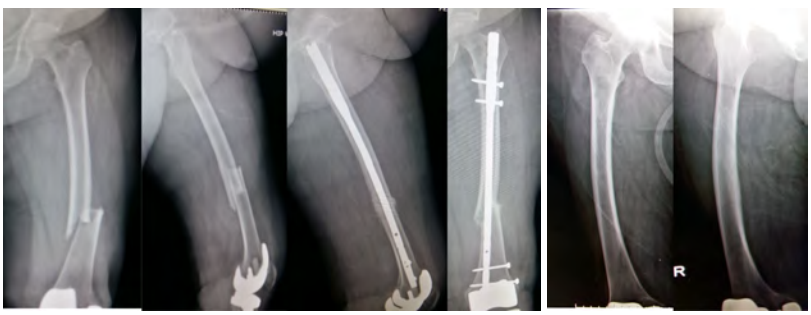

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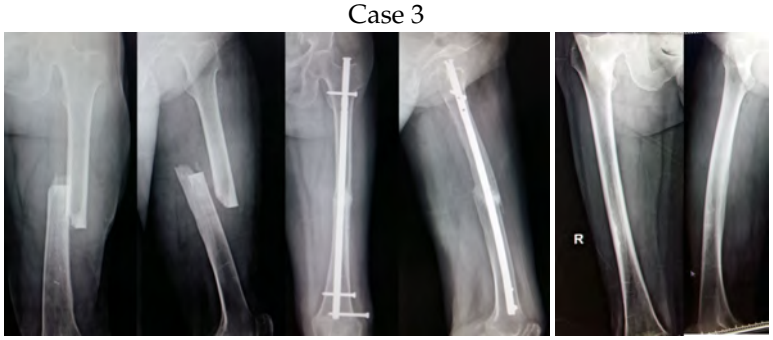

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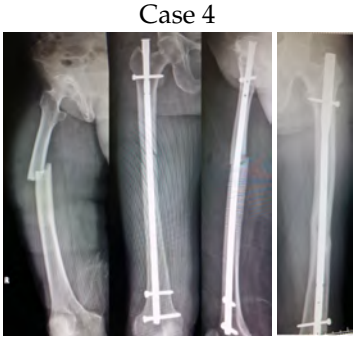

Case 5

Figures 1–5. Pre op X-ray showing radiological features, post op X-ray showing united fracture and X-ray of contralateral femur.

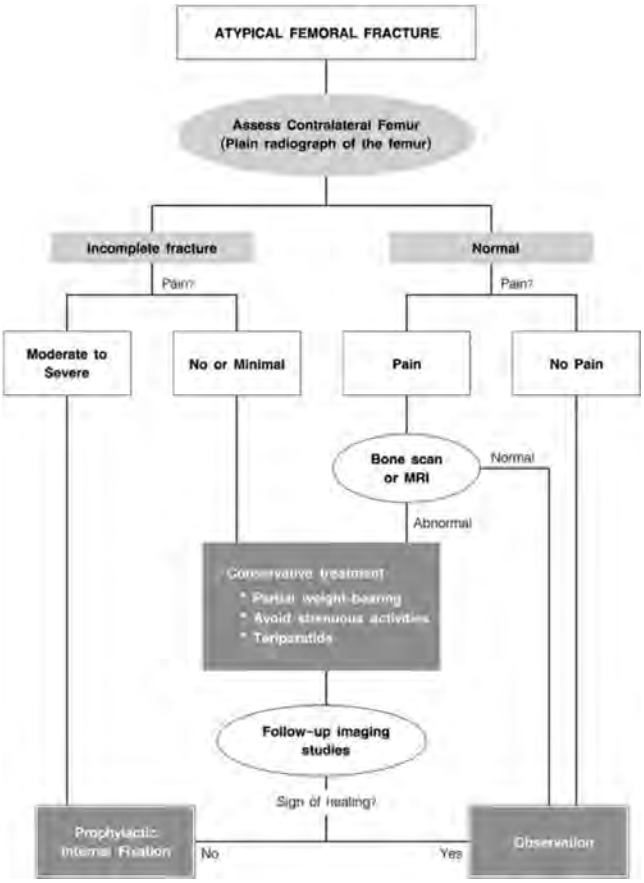

Figure 6. Assessing Contralateral femur in atypical fracture.

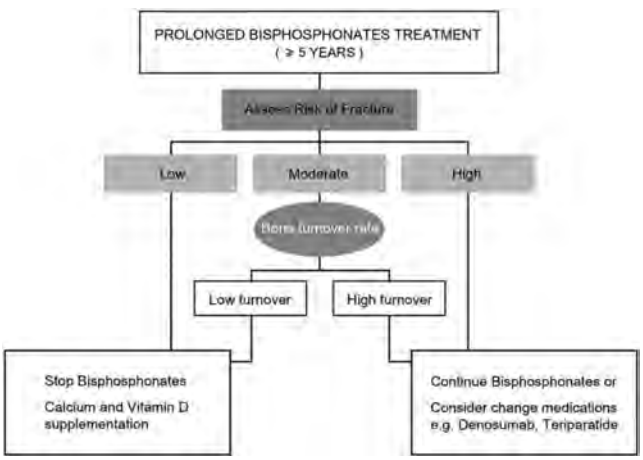

Figure 7. Management of Patients On bisphosphonates with atypical fracture.

be corrected to a minimum of 30 ng/mL.[9] Teriparatide should be initiated postoperatively to increase bone turnover and BMD in patients previously taking bisphosphonates. Surveillance of patients with atypical femoral fractures throughout the healing process is essential because bilaterality is a minor feature of these fractures, and incipient stress fractures in the contralateral limb may propagate when weight bearing is reduced in the fractured limb. Radiographs of the contralateral femur must be evaluated for evidence of a stress fracture. A technetium bone scan or magnetic resonance image (MRI) should be considered if a stress fracture is suspected. Prophylactic intramedullary nail fixation should be considered when there is a moderate to severe pain of the affected limb, persistent pain or persistence of the fracture line on radiographs after 3 months of conservative treatment, or progression of the fracture line observed from serial radiographic examination (Figure 6).

For patients on bisphosphonates for durations longer than 5 years, the FDA currently recommends periodic review of the medical management of patients on long-term bisphosphonate therapy (Figure 7), although the elements of those reviews are still under debate. These patients should be monitored for signs of femoral stress fractures as well as for markers of bone turnover to avoid over suppression of remodeling. Relevant markers include serum or urine N- or C-telopeptide of collagen cross-links (NTX, CTX) for bone resorption and bone specific alkaline phosphatase or aminoterminal propeptide of type I collagen (PINP) for bone formation.[10] In addition, patients should be monitored for evidence of stress fractures, particularly prodromal pain in the thigh or groin, because bisphosphonates bind preferentially to sites of microdamage formation, and localized over suppression of bone turnover and damage accumulation could potentially occur even with normal systemic levels of bone turnover markers. Finally, fracture risk should be assessed by considering individual patient risk factors such as age, history of glucocorticoid use, and family history of fragility fracture.

Summary

The absolute risk of atypical femur fracture is low compared to the risk of typical femur fractures in bisphosphonate users, but current epidemiologic evidence suggests that long-term bisphosphonate use may be an important risk factor for atypical fractures. Long-term bisphosphonate users must be monitored for biochemical markers of bone turnover and evidence of stress fractures in the proximal femur.[10,11] In patients with atypical femoral fractures, bisphosphonates should be discontinued, and teriparatide should be initiated postoperatively to increase bone turnover and bone density. There is no rationale for withholding bisphosphonates therapy from patients with osteoporosis, although continued use of bisphosphonates beyond a treatment period of 3 to 5 years should be revaluated

annually. Consideration should be given to stopping (at least temporarily) bisphosphonates therapy in patients who are reassessed to be at low or low-moderate risk (no incident fractures, T-score of 2.0 or higher, and no other major risk factors) after a 3 to 5 years therapeutic period.[11]

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