Case report

Intertrochanteric fracture associated with undiagnosed ipsilateral incomplete atypical femoral shaft fracture without bisphosphonate use: Unique case report and literature review.

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1. Introduction

Atypical femur fractures (AFFs) are often associated with bisphosphonate (BP) administration for postmenopausal or senile osteoporosis,^{1,2} while reports on AFFs without prior history of bisphosphonate use are limited.^{3,4} However, recently (2013) the definition of AFFs does not mention prior BP administration.¹ AFFs occur as stress/insufficiency fractures and can result from significant femoral bowing.^{3–5}

This case report presents an incidentally diagnosed atypical femoral shaft fracture (AFSF) ipsilaterally to a traumatic intertrochanteric fracture.

To our knowledge, this is the first report of an AFSF in an elderly patient with no prior history of BP treatment for osteoporosis and a concomitant intertrochanteric fracture.

2. Case report

An 87-year-old female patient with no history of previous trauma was admitted to our emergency department for severe pain in her left hip while walking in the last two days. She had undergone an intramedullary nailing in her left hip for an intertrochanteric fracture at another institution one month before.

Postoperatively, she was successfully mobilized on two crutches, with mild pain in her left hip and thigh. Two days before admission to our institution, she experienced severe pain in her hip and could no longer bear weight. The patient had never been subjected to any antiosteoporotic treatment. Physical examination revealed pain during passive motion in her left hip, with no local edema, redness, or warmth at the scar site, which would have indicated infection from the recent surgery. Her blood counts analysis was within normal limits: WBC: 8.9 k/mL (normal: 4–10.0 k/mL), CRP: 0.1 mg/dL (< 0.3 mg/dL), ESR: 14 mm/1 st h (0–20 mm/1 st h), and GFR value: 63 mL/min/1.73 m². On admission, the roentgenographic examination detected a cranial cut-out of the lag screw associated with an ipsilateral incomplete atypical femoral fracture with transverse configuration and concomitant lateral cortex hypertrophy at the nail tip (Fig. 1a, b).

We immediately thought this femoral fracture could be related to stress-concentration at the distal nail tip. However, while we were searching for the initial pre-operative radiographs, the patient reported having had a mild thigh pain over the last three months, which she thought was typical osteoarthritic hip pain. Then, the initial plain roentgenograms, once found, revealed a small, barely noticeable transverse radiolucency line (Fig. 2a, b).

Coincidentally, surgeons had implanted a short nail that ended at the exact area of the atypical shaft fracture (Fig. 3). Authors replaced the short nail with a long cephalomedullary nail to address both lesions (Fig. 4a, b). The patient's metabolic profile {TSH: 2.1 µIU/mL (normal limits 0.27–4.2), PTH: 60 pg/mL (normal limits 15–65), ALP: 52 IU/l (normal limits 50–136), calcium: 8.3 mg/dL (normal limits 8.5–10.1), phosphorus: 3.5 mg/dL (normal limits 3–4.5), cortisol: 13.1 µg/dL (normal limits 6–23), aldosterone: 75 pg/mL (normal limits 10–160), albumin: 3.1 (normal

Abbreviations: AFFs, atypical femur fractures; AFSF, atypical femoral shaft fracture; CRP, C-reactive protein; ESR, erythrocyte sedimentation rate; WBC, white blood cells; TSH, thyroid stimulating hormone; PTH, parathyroid stimulating hormone; ALP, alkaline phosphatase; NF-kB, nuclear factor –kappa-B; GFR, glomerular filtration rate; DEXA, dual-energy x-ray absorptiometry.

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Fig. 1. a,b: Profile and anteroposterior hip radiogram showing the cut out of the lag screw with concomitant atypical femoral fracture at the tip of the nail.

Fig. 2. a,b: AP and profile hip radiogram from our patient initial admission, showing the intertrochanteric and the ipsilateral atypical femoral fracture.

Fig. 3. AP proximal femur radiogram after the initial hip surgery.

Fig. 4. a,b: AP hip and femur radiogram after long gamma nail implantation and a focused radiogram highlighting the stress fracture of our case.

Fig. 5. AP hip and femur radiogram measuring the coronal femoral bowing of our case.

limits 3,4–5,0)) revealed no evidence of secondary osteoporosis, although we noticed a mild 25-OH-vitamin D deficiency {9,5 ng/mL (normal limits 11,10–42,90). Unfortunately, the patient had never received a DEXA scan to assess her bone density. Before discharge from the author's department, teriparatide injections plus calcium and vitamin D were prescribed to accelerate bone healing.

3. Discussion

The reported risk of AFFs in adult patients receiving BPs for osteoporosis ranges from 0.006% to 1.1%.^{1,6} A longer duration of bisphosphonate use is considered a risk factor for AFFs.^{1,2,6} If bisphosphonates have been administered for less than two years, the AFF incidence is approximately 2 in 100,000, while bisphosphonate use for eight or more years is associated with an AFF incidence of approximately 78 in 100,000.⁶ Few studies have reported AFFs without bisphosphonate administration^{3,4,6}; thus, in 2013, the American Society for Bone and Mineral Research revised the definition of AFFs by deleting the clause pertaining to bisphosphonate use. The five major features of the current definition of AFFs are: subtrochanteric or diaphyseal location, medial spike or lateral cortex hypertrophy, transverse or short oblique configuration, no comminution, and minimal or no association with trauma.¹

Knowledge is limited regarding atypical femur fractures with no history of BPs uptake, although, in case series, incidence of these fractures ranges from 8% to 22% of all AFFs.^{3,4,6} The important point in the pathogenesis of AFFs is lateral cortex failure due to increased tensile stress.⁵ Thus, AFFs have all the characteristics of a stress fracture, and increased tensile stress is the critical point in AFFs pathogenesis, both with and without BPs use. Stress fractures heal by targeted remodeling of the injury site through osteocyte apoptosis, which signals for repair by increased production of the receptor activator of NF- κ B ligand, osteoclastic resorption to remove the damage, and osteoblastic formation to replace resorbed bone.¹ The increased incidence of these fractures in BP users can be explained by the deactivated osteoclasts and the suppressed remodeling process that accompany BP use, which also adversely affects intracortical repair of the developing stress fracture in AFFs, allowing cracks to grow to critical size. For these reasons, bisphosphonates should be discontinued or not prescribed in AFF patients.¹

AFF incidence and fracture location are strongly correlated with the degree of femoral bowing.^{3,5} Femoral bowing is measured as the angulation between the proximal and distal quarters of the femoral diaphysis with a cut-off value of $>5.25^\circ$.⁵ In our case, the lateral bowing was 12° (Fig. 5). AFFs are generally associated with delayed union, which can occur regardless of BP intake.⁴ Most of these fractures are recognized at the diaphyseal region of the femur when

not associated with BP medication,⁴ whereas diaphyseal AFFs are correlated with larger lateral and anterior bowing.^{3,5}

The decision whether to operatively treat incomplete atypical femoral fractures is controversial. The literature suggests prophylactic nailing,⁷ because it is easier on incomplete fractures, and healing and hospitalization times are shorter. The quality of the reduction of these fractures appears to accelerate bone union and decrease healing time.⁸ Thus, early diagnosis and treatment of incomplete AFFs provides a good opportunity for excellent results. When these incomplete fractures are accompanied by significant bowing, surgeons should consider the mismatch between implant design and femur curvature, so as to avoid intraoperative complications, such as femoral shaft comminution.⁷ Sick elderly patients with several comorbidities may have only one opportunity to receive peritrochanteric fracture repair. Inappropriate implant selection must be avoided because failure and revision surgery can result in disastrous complications.⁹ Based on the current literature, teriparatide appears to be the best medication for treating osteoporosis accompanied by an AFF. This anabolic agent often enhances fracture healing.¹⁰

4. Conclusion

Knowledge of AFF pathogenesis, good quality radiograms of the entire femur including the hip and knee joint in all elderly patients with peritrochanteric fractures, accompanied with high degree of suspicion are needed to recognize this combined injury. Furthermore, medical history of pre-existing thigh pain is useful, while history of bisphosphonate use is unnecessary for documenting an AFF diagnosis. Identifying this rare entity differentiates the treatment choice and strongly affects the outcome, and this case report provides a clear example. Femoral bowing affects the AFF incidence and location regardless of bisphosphonate administration. Patients with intertrochanteric fracture and

increased femoral bowing could be treated using long nails while avoiding BP use, although more studies are needed to confirm and document the cost-effectiveness of this method.

Conflict of interest

None.

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