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Case Report

Non-traumatic Atypical Peri-implant Femoral Fracture at the Distal Screw after Short Femoral Nail Fixation for a Pertrochanteric Fracture

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This is the second report of an atypical peri-implant femoral fracture occurring at the distal screw after intramedullary nail fixation of a pertrochanteric fracture. A 94-year-old Japanese female with a 5-year history of alendronate intake presented with prodromal pain in her right thigh after intramedullary nail fixation. Plain radiographs showed an incomplete noncomminuted transverse fracture at the distal screw, suggesting an atypical peri-implant femoral fracture. The pathogenesis of an atypical peri-implant femoral fracture could be a combined systemic bone metabolism disorder and repetitive overloading at the screw, similar to the pathogenesis of an atypical periprosthetic femoral fracture around stem implantation.

Key words: atypical femoral fracture, insufficiency fracture, bisphosphonate, atypical peri-implant femoral fracture, atypical periprosthetic femoral fracture

Atypical femoral fracture (AFF) has recently been defined and is becoming comparatively well-known [1]. Although the current definition of AFF by the American Society for Bone and Mineral Research (ASBMR) excludes periprosthetic fractures [1], several authors have reported atypical periprosthetic femoral fractures around the stem in response to low-energy trauma which fulfill the major criteria for AFF, and these fractures were radiographically identical to AFFs [2-4].

Case reports of atypical peri-implant femoral fractures have begun to appear in the literature, describing patients presenting with impending or complete peri-implant fractures that are radiographically identical to AFF. The occurrence of an atypical peri-implant femoral fracture after internal fixation (such as fixation with an intramedullary [IM] nail or plating) for hip fractures is extremely rare compared to the occurrence of atypical periprosthetic femoral fractures. To our knowledge, there are only 7 reports of a non-traumatic atypical peri-implant femoral fracture that occurred around the implant [5-9], including one case at the distal screw [5].

Herein we present a case of a non-traumatic peri-implant femoral fracture that progressed just at the distal screw after a repair using a short femoral nail for a pertrochanteric fracture; this factor was presumably caused by severe osteoporosis, suppressed bone metabolism by long-term bisphosphonate (BP) use, and stress concentration at the distal screw. We also review the pertinent literature of non-traumatic atypical peri-implant femoral fracture, with a focus on its pathogenesis.

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Case Report

A 91-year-old Japanese woman with a 2-year history of alendronate intake presented with right pubic pain that occurred during normal daily activities, without falls. Her body weight was 40 kg and her height was 142 cm. She could walk slowly using a walking frame. Her medical history included hypertension. She did not take any other medication, including glucocorticoids or proton pump inhibitors. Plain radiographs showed no clear fracture line, but there was a slight periosteal thickening at the lateral cortex of the right femur (Fig. 1).

Magnetic resonance imaging (MRI) of the pelvis demonstrated an acute right pubic fracture. MR images of the femurs demonstrated focal low intensity on T1-weighted images and high intensity on short tau inversion recovery (STIR) images only of the right femur (Fig. 1), suggesting bone edema, although the patient had no thigh pain. The T-scores at the total hip and lumbar spine were −4.6 and −3.8, respectively, as determined by dual X-ray absorptiometry. The laboratory blood test results were within normal ranges. She was diagnosed with a right pubic fracture. Bone union was achieved with conservative treatment after 2 months, but with residual malunion.

At age 93 years, the patient presented with right hip pain after falling from a standing height. Radiographs showed a right pertrochanteric fracture (Fig. 2), which was AO/OTA 31A2.2 according to the AO/Orthopaedic Trauma Association classification [10]. The appearance of flaring was clearer than it was 2 years earlier. Repair of the pertrochanteric fracture was performed with a cephalomedullary nail (Gamma 3, Stryker Trauma, Kiel, Germany; 12 mm diameter, 170 mm length, 125° neck angle). The distal static screw was inserted one time smoothly without accidental additional drill holes. The fracture fragment was reduced to within the acceptable range. Bone union was achieved at 3 months postoperatively without displacement (Fig. 2). The patient was able to walk slowly with a walking frame and without pain.

At age 94 years, she presented with right thigh pain that had gradually increased during normal activity. She reported a 5-year history of alendronate intake. She had no history of trauma in the past year. Plain radiographs showed no fracture line. Radiographs obtained 2 months later showed an incomplete noncomminuted

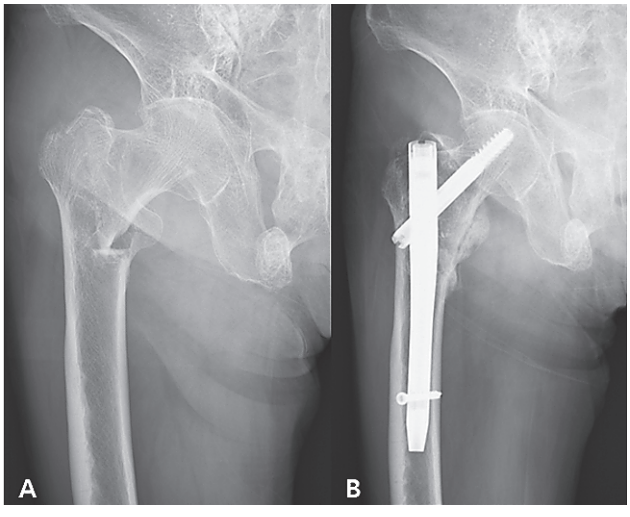

Fig. 2 A, Right pertrochanteric fracture; B, Bone union was achieved 3 months after internal fixation with a cephalomedullary nail.

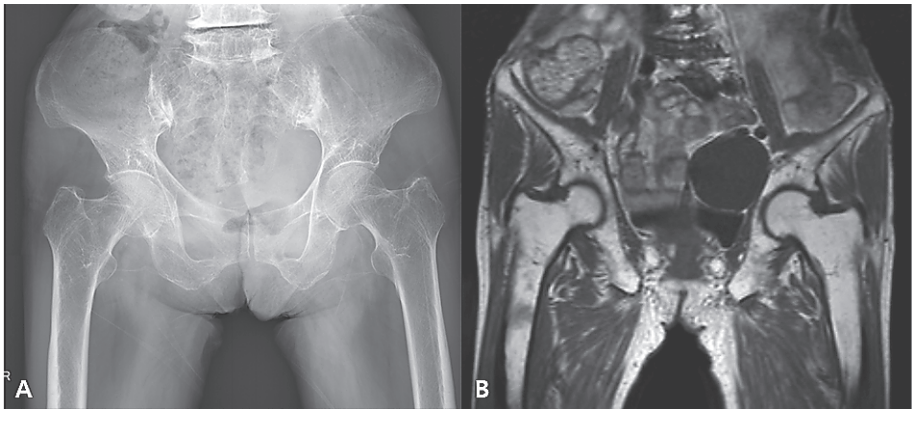

Fig. 1 A, Plain radiograph showed no clear fracture line on the right femur and a slight periosteal thickening of the lateral cortex; B, Focal low intensity on T1-weighted image at the right femoral shaft.

transverse fracture located anterior to the distal screw (Fig. 3). The fracture line was similar to the "dreaded black line" anteriorly, indicating a tension fracture caused by bony resorption. The contralateral left femoral bone was also osteoporotic and similar in regard to the periosteal thickening of the lateral cortex. Regarding the right femur, the neck-shaft angle was 128°. The femoral curvature, measured as the angles between 2 linear lines drawn along the proximal and distal portions of the femoral shaft [11], was 8° for the anteroposterior view and 14° for the lateral view. There was no apparent implant loosening, toggling motion, or malalignment of the nail components.

The fracture fulfilled 4 of the 5 major features for AFF—namely, it was not associated with trauma, it was a complete transverse fracture at the lateral cortex, and it was noncomminuted—and 2 of 4 minor features, *i.e.*, a generalized increase in the cortical thickness of the femoral diaphysis and unilateral prodromal symptom, a although periprosthetic fracture is excluded from the current definition [1]. Based on these findings, we diagnosed the patient with a peri-implant femoral insufficiency fracture or an atypical peri-implant femoral fracture related to her BP use. Osteoporosis, abnormal signal intensity on previous MR images, and gradual accumulation of microcracks around the distal screw area at the overload site were thought to be involved in this atypical peri-implant femoral fracture. Operative treatment—such as exchanging the long nail or removing the distal screw—was recommended to prevent a subsequent complete fracture, but the patient decided to continue conservative treatment (*i.e.*, cessation of alendronate use). She was later prescribed teriparatide and was restricted to limited weight-bearing depending on the presence and degree of thigh pain.

At age 95, 6 months after the fracture at the distal screw site, the patient presented with persistent pain in her right thigh. The fracture line was clearer at the lateral and anterior cortex than on previous scans. She chose to have minimally invasive surgery, and thus distal screw removal was performed. Plain radiographs were taken 1 year after the screw removal, which showed that the callus was bridging gradually, but no bone union was seen.

When the patient reached 98 years old (3 years after the screw removal), bony union was achieved, suggesting delayed fracture healing (which is one of the minor features of AFF) (Fig. 4). She had no pain and regained the ability to walk with a walking frame, although her activity was limited by her comorbidities.

The patient was informed that data from the case would be submitted for publication, and she gave consent.

Discussion

In this case, a non-traumatic atypical peri-implant femoral fracture was seen only at the distal screw site (detected by abnormal intensity on previous MR images) after a short femoral nail was inserted; the fracture was presumably caused by severe osteoporosis, suppressed bone metabolism (from long-term BP use), and a concentration of stress after implant fixation. This fracture

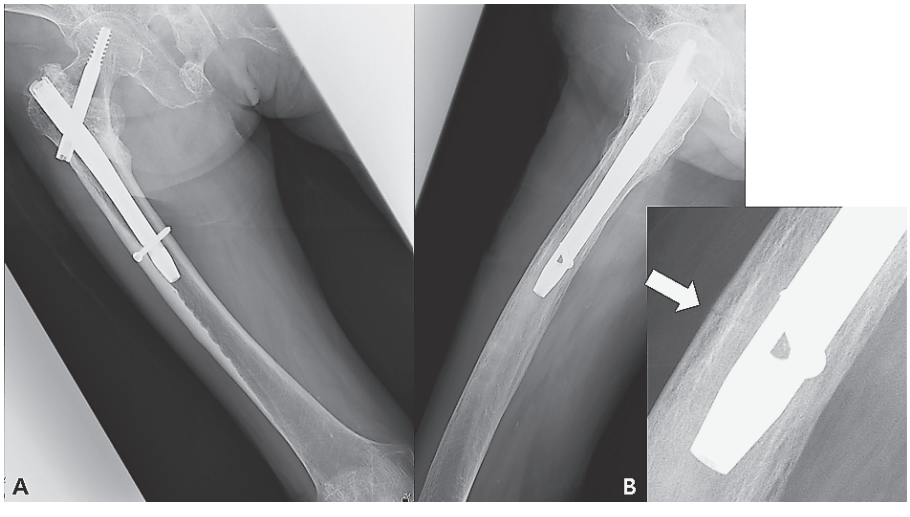

Fig. 3 **A**, Anteroposterior view on plain radiograph showed lateral bowing and no fracture line; **B**, Lateral view on plain radiograph showed an incomplete noncomminuted transverse fracture located anterior to the distal screw (arrow).

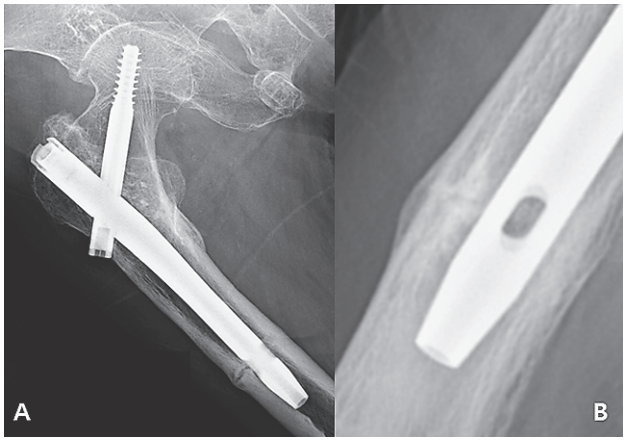

Fig. 4 A, B, Three years after the distal screw removal, bony union was achieved.

was pathophysiologically identical to an insufficiency fracture and was radiographically identical to AFF. Our patient's case thus represents a new problem: an atypical peri-implant femoral fracture after bone union was achieved following internal fixation for a hip fracture.

Etiology. Atypical peri-implant femoral fracture is a recent concept. A non-traumatic peri-implant femoral insufficiency fracture is defined as a fracture occurring during normal activity or physiological stress in day-to-day activities, and it is extremely rare. Our review of the literature identified only seven such fractures [5-9] (Table 1). Patients with any trauma, including falling [12, 13], malignancy [3], infection, or neurological deficit, were excluded from our literature search.

All of the previously reported patients [5-9] were female and had osteoporosis. The mean duration of BP use, *i.e.*, alendronate, was 6.4 years in 6 of the 7 patients. These patients had experienced prodromal thigh pain for several months, and radiographs showed thickening of the cortex and a transverse fracture line. A thickened and beaked lateral cortex may be an early sign of lateral cortical hypertrophy (as is seen for AFF), which is not at all like the presentation of a typical traumatic peri-implant fracture. Following the bone union of the patients' primary fractures, the mean time to secondary insufficiency fractures was approximately 4.9 years. The method of internal fixation for the primary fracture was a plate for 4 fractures and an IM nail for four fractures, although the stress riser effect is speculated to be lesser with nails than with plates in the diaphysis of the femur.

Our patient's case is only the second case of an atypical fracture at the distal screw site after fixation with a short cephalomedullary nail [5]. The reported patients with a diaphyseal AFF were older and had a lower body mass index, lower low bone mineral density (BMD), and larger lateral and anterior bowing than those with a subtrochanteric AFF [14]. These characteristics of patients with atypical peri-implant femoral fractures are similar to those of patients with diaphyseal AFF. We speculate that atypical peri-implant femoral fractures may have a pathology similar to that of atypical periprosthetic femoral fractures.

Pathogenesis. The exact cause of these fractures is unknown and likely multifactorial. We focused on 2 points, *i.e.*, BP use (a biological factor) and anterolateral bowing and altered stress distribution after implant fixation (mechanical factors). Because BPs localize in areas that are already developing stress fractures, the suppression of targeted intracortical remodeling at the site of an AFF is likely to impair the processes by which stress fractures normally heal [1]. A meta-analysis reported that BP exposure was associated with an increased risk of femoral subtrochanteric fracture, femoral shaft fracture, and AFF [15].

The lateral cortex of the femur is known to sustain high levels of tensile stress due to bending force [16]; therefore, people with lower limb geometry that could exacerbate that effect (*e.g.*, a bowed femur, or Asian race) could be predisposed to damage in this location [1, 11, 17]. A mechanical analysis demonstrated that significant tensile stress on the anterolateral surface of the femur caused by bowing deformity can induce an AFF in the femoral diaphysis [18]. With a greater lateral bowing angle, the AFF location was moved from the subtrochanteric area to the diaphysis area [14]. A multivariate analysis revealed that age >65 years and low BMD were related to AFF in the diaphysis [14].

Implants concentrate stress at the plate-bone junctions due to differential stiffness. Dynamic strains under tension may be accentuated, contributing to local microdamage that overwhelms the impaired healing capacity of the antiresorptive-treated bone. These areas can then become more susceptible to chronic stress reactions and subsequent atypical fractures [12], despite increased cortical thickness [8]. In our patient's case, the distribution of tensile strain moved to the distal screw area after fixation with an IM nail [19]; repeated tensile stress accumulation at this site might be the cause of the insufficiency fracture of the severely

Table 1 Literature review findings of cases with non-traumatic atypical peri-implant femoral fracture

Authors	Bjorgul *et al*. [9]	Cho *et al*. [6]	Kim *et al*. [7], right side	Kim *et al*. [7], left side	Yoon *et al*. [10]	Kumar *et al*. [8], case 1	Kumar *et al*. [8], case 2	Present case
Country	Norway	Korea	Korea	Korea	Korea	United Kingdom	United Kingdom	Japan
Age (years)/Sex	73/F	66/F	81/F	76/F	74/F	57/F	56/F	94/F
Height (cm)/ Weight (kg)	NA	NA	NA	NA	150/45	NA	NA	142/40
Co-existing illness	OP, AUF at 82 y.o.	OP	OP	OP	OP	OP	OP, Chronic renal failure	OP, hypertension fragility fracture of the pelvis
Medication	Alendronate	Alendronate	NA	NA	Alendronate	Alendronate	Alendronate	Alendronate
Duration of bisphosphonate (years)	7	14	NA	NA	3.5	4	5	5
T score	NA	NA	−3.6 at lumbar spine	−3.6 at lumbar spine	−4.1	NA	NA	−4.6 at total hip, −3.8 at lumbar spine
Previous fracture	Left pertrochanteric fracture	Right intertrochanteric fracture	Right AFF	Left AFF	Right pertrochanteric fracture	Left femoral neck fracture	Left intertrochanteric fracture	Right pertrochanteric fracture
Previous internal fixation	Sliding hip screw	Sliding hip screw	Antegrade long IMN	Antegrade long IMN	Short CMN	Sliding hip screw	Sliding hip screw	Short CMN
Time elapsed from the previous fracture to atypical peri-implant femoral fracture (years)	7	14	2	3	3.5	4	NA	1
Location of atypical peri-implant femoral fracture	between 2nd and 3rd screw from most distal	below the plate end	distal one of the proximal screws	distal one of the proximal screws	distal screw	below the plate end	below the plate end	distal screw
Type of atypical peri-implant femoral fracture	complete transverse at lateral, spike at medial	complete transverse at lateral thickened cortex	incomplete at anterior thickened cortex	incomplete at anterior thickened cortex	focal eccentric sclerosis of the lateral cortex, thinning of the lateral femoral cortex	incomplete at lateral	incomplete at lateral	incomplete t ransverse at lateral thickened cortex
Treatment	implant removal, antegrade long IMN	implant removal, antegrade long IMN	conservative treatment	conservative treatment	implant removal, antegrade long IMN	implant removal, antegrade long IMN	distal screw removal, long retrograde femoral nail	distal screw removal
Follow-up period (months)	60	12	60	96	12	18	24	48
Outcome	bone union	bone union	delayed union	delayed union	bone union	bone union	bone union	bone union

F, female; OP, osteoporosis; AUF, atypical ulnar fracture; AFF, Atypical femoral fracture; IMN, intramedullary nail; CMN, cephalomedullary nail; NA, not available.

osteoporotic bone.

After plate fixation, stress is usually concentrated at the transitional zone from the plate to the bony cortex due to the difference in structural stiffness between the normal bone and the plated bone segment [20]. Some affecting factors include stress shielding, altered stiffness at the screw of the plates [21], and decreased blood supply in the cortical bone adjacent to the plate. In contrast, an IM nail was reported to be able to reduce external mechanical factors, as this type of nail provides a more homogenous distribution of mechanical forces compared to plate fixation [13]. However, in the presence of osteoporosis, this toggle motion is higher and allows rotation of the femur around the nail under physiological loading [22]. Greater stress is caused at the tip of the nail, which may lead to a higher incidence of postoperative secondary femoral shaft fractures. We speculate that the pattern of the tensile strain changes or tensile strain accumulates at the screw area after fixation, regardless of whether a nail or a plate is used; repeated micromotion might cause insufficiency fractures in the severely osteoporotic bone [7].

Diagnosis. Orthopedic surgeons should be aware of atypical peri-implant femoral fractures. Close monitoring with repeated radiographs over time may be necessary for an accurate diagnosis, because clinical symptoms of incomplete fracture appear insidiously and radiographic findings are absent or subtle in the early stages. In our patient's case, although the fracture was transverse, it was detected at the anterior cortex, due to the difficulty in detecting transverse fractures at the lateral cortex after a screw has been inserted at the lateral cortex [7]. To better understand the rare characteristics of atypical peri-implant femoral fractures, physicians should pay attention to their patients' recent history of BP treatment, hip fracture surgical treatment, total femur geometry, and subtle changes detected with sequential images. When plain radiographs show a suspicious fracture line, bone scintigraphy is useful to obtain more information on the osteoblastic condition at the stress riser area [5,6]. In addition, each patient's bone metabolism and secondary osteoporosis must be assessed.

Treatment. Simple surgical fixation of the fractured bone without optimizing other predisposing biological and mechanical factors may not prevent the progression of subsequent atypical peri-implant femoral fractures. Specific medical treatments should be performed for systemic bone metabolism, especially in patients with long-term BP use, and correct surgical treatment should be administered for the local fracture bone while considering the stress distribution after implant fixation.

The ASBMR task force recommended that the following may prevent AFF: the discontinuation of BPs, adequate calcium and vitamin D, and the administration of teriparatide [1]. Teriparatide is used to treat osteoporosis and to promote fracture healing.

Screws should not be inserted at the inflection point of bowing or in areas with an abnormal sign on MRI in previous images. If an incomplete fracture causes moderate-to-severe pain, it is reasonable to consider prophylactic internal fixation [23]. An IM nail of sufficient length and diameter is recommended to reduce the risk of another stress fracture in the area where the load is concentrated [7]. According to Koch's model [24], the forces along the lateral aspect of the femur taper distally from the highly tensile subtrochanteric region, becoming compressive along the distal lateral metaphysis. Cadaveric models have demonstrated that longer IM nails distribute strain throughout the distal femur, transmitting progressively decreasing loads to the proximal femur [25]. Therefore, ending an IM nail in the lower metaphyseal region may reduce the risk of sustaining implant-related atypical fractures [12].

Avoiding distal screw placement is a key treatment option. Secondary traumatic femoral fractures can occur if relatively large screws are used for small femur bones [26]. Pertrochanteric fractures (31-A1,2) can be treated successfully with IM nails without distal screws [27]. As described in our patient's case, the use of a long IM nail without distal screws might be a possible fixation strategy for a pertrochanteric fracture, *i.e.*, a suspected atypical femoral shaft fracture.

We believe that the recognition of an atypical peri-implant femoral fracture would be the first step toward further investigation. For the next case of an atypical peri-implant femoral fracture, a bone turnover biochemical marker, bone pathological testing, and bone histomorphometric testing should be used to confirm the patient's bone metabolism and activity. Such a case may call for the introduction of an atypical peri-implant femoral fracture as its own entity or a revision of the definition of AFF, given that these peri-prosthetic/peri-implant fractures appear to be similar to AFF in terms of etiology [3,12].

In conclusion, physicians should be aware that an atypical peri-implant femoral fracture can develop due to the combined effects of stress concentrations caused by previously fixed implants and skeletal fragility due to osteoporosis or the long-term use of BPs. Patients with prodromal thigh pain after their hip fracture has healed following repair with an implant require close observation for the possible development of an atypical peri-implant femoral fracture.

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