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Case Report

A secondary atypical diaphyseal femoral fracture after intramedullary nailing for an atypical subtrochanteric femoral fracture: A case report

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ABSTRACT

Intramedullary nailing is the gold standard of treatment for atypical femoral fractures, with a few reports of secondary atypical subtrochanteric femoral fractures following intramedullary nailing for atypical diaphyseal femoral fractures. However, there are no reports of secondary atypical diaphyseal femoral fractures following intramedullary nailing for atypical subtrochanteric femoral fractures.

A 71-year-old woman with adult-onset Still's disease sustained a right atypical subtrochanteric femoral fracture and was treated with a mid-length intramedullary nail. One year after the surgery, the patient sustained a contralateral atypical diaphyseal femoral fracture and was treated with a long-length intramedullary nail. Moreover, 6 months after the second surgery, the patient complained of right-thigh pain, and a radiograph of the lateral view of the femur revealed a diaphyseal femoral fracture at the distal screw-insertion site. Revision surgery was performed using a long-length nail and screws directed toward the femoral head. Bony union of the bilateral diaphyseal femoral lesion was obtained, but the subtrochanteric lesion remained unhealed 1.5 years postoperatively.

Mid-length intramedullary nailing for atypical subtrochanteric femoral fractures can cause secondary atypical diaphyseal fractures because of stress concentration at the distal screw-insertion site. For atypical subtrochanteric femoral fractures, the use of long-length nails and proximal screws directed toward the femoral head may be important to prevent secondary atypical diaphyseal femoral fractures.

Introduction

Atypical femoral fractures are often associated with long-term use of bisphosphonates [1]. However, recent studies have shown that multiple factors contribute to atypical femoral fractures, which include not only drugs like bisphosphonates, glucocorticoids, and proton pump inhibitors, but also race, genetic mutations, femoral curvature, femoral neck-shaft angle, lower extremity alignment, and loading stress [1,2]. Treating complete fractures is challenging for orthopaedic surgeons because of their high revision rates compared to typical femoral fractures [1].

The first-line treatment for atypical diaphyseal or subtrochanteric femoral fractures is intramedullary nailing. Recently, secondary

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subtrochanteric femoral fractures have been reported after intramedullary nailing for atypical diaphyseal femoral fractures with minor or no traumatic episodes [3–6]. However, there have been no reports of diaphyseal femoral fractures following intramedullary nail treatment for atypical subtrochanteric femoral fractures.

Herein, we present a case of an atypical diaphyseal femoral fracture after intramedullary nailing for an atypical subtrochanteric femoral fracture.

Case presentation

A 71-year-old woman with adult-onset Still's disease was admitted to the emergency department of another hospital after falling from standing height. She had been taking bisphosphonates once a month and glucocorticoids daily for 5 years. Upon admission, the patient complained of severe right-thigh pain and deformity. Plain radiographs revealed a right subtrochanteric femoral fracture (Fig. 1A), and based on the fracture findings, location and configuration of the fracture, medial spike, and history of trauma, the patient was diagnosed with an atypical subtrochanteric femoral fracture [7].

Surgery was performed on the day after the injury, and a mid-length nail was inserted after open reduction and provisional fixation using a titanium cable (Fig. 1B). Postoperatively, the patient was allowed full weight-bearing as tolerated, bisphosphonates were discontinued, and parathyroid hormone was administered. On radiography, the varus deformity of the proximal fragment gradually progressed and bony union was not observed. However, the patient did not complain of pain. One year after the initial surgery, the patient was brought to our emergency department complaining of severe left-thigh pain and deformity. Plain radiographs showed a left diaphyseal femoral fracture (Fig. 2A), which was diagnosed as an atypical diaphyseal femoral fracture because the features of this fracture were the same as those of the contralateral atypical subtrochanteric femoral fracture. The surgery was performed on the day following the injury; a long-length nail was used after closed reduction, and proximal screws were inserted toward the femoral head (Fig. 2B).

Six months after the left femoral fixation, the patient complained of right-thigh pain and visited our outpatient clinic. An anteroposterior view of the radiograph showed no abnormal findings, but a fracture line was suspected at the anterior part of the distal screw on a lateral view of the radiograph (Fig. 3A). Computed tomography confirmed this finding and revealed an incomplete transverse fracture around the distal screw-insertion site (Fig. 3B). To prevent a complete fracture the patient underwent revision surgery for the right incomplete atypical femoral diaphyseal fracture. The preexisting mid-length nail was removed, and a new long-length nail was inserted after realignment and provisional plate fixation of the subtrochanteric lesion (Fig. 4A).

One and a half years after the revision surgery, bony union of the diaphyseal lesions in the right and left femurs was observed (Fig. 4B and C). Although the bony union of the subtrochanteric lesion in the right femur had not been achieved at the time, the patient could walk without pain.

Discussion

We herein report a case of secondary atypical diaphyseal femoral fracture after mid-length intramedullary nailing for an atypical subtrochanteric femoral fracture.

Kim et al. [6] reported a case series of secondary subtrochanteric or diaphyseal femoral fractures after atypical femoral fracture fixation using nails or plates, which they termed as peri-implant atypical femoral fractures. According to previous reports [3–6], peri-

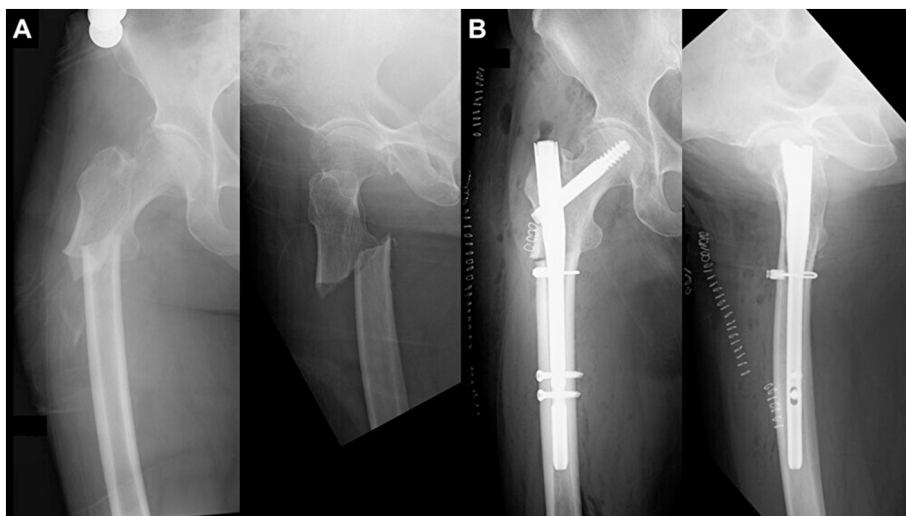


Fig. 1. Pre- and post-operative radiographs of the right femur. (A) Plain radiographs show an atypical subtrochanteric femoral fracture with a transverse fracture line and medial spike. (B) The right femur is fixed using a mid-length intramedullary nail.



Fig. 2. Pre- and post-operative radiographs of the left femur. (A) Plain radiographs show an atypical diaphyseal femoral fracture with the same features as the right femur. (B) The left femur is fixed using a long-length intramedullary nail.

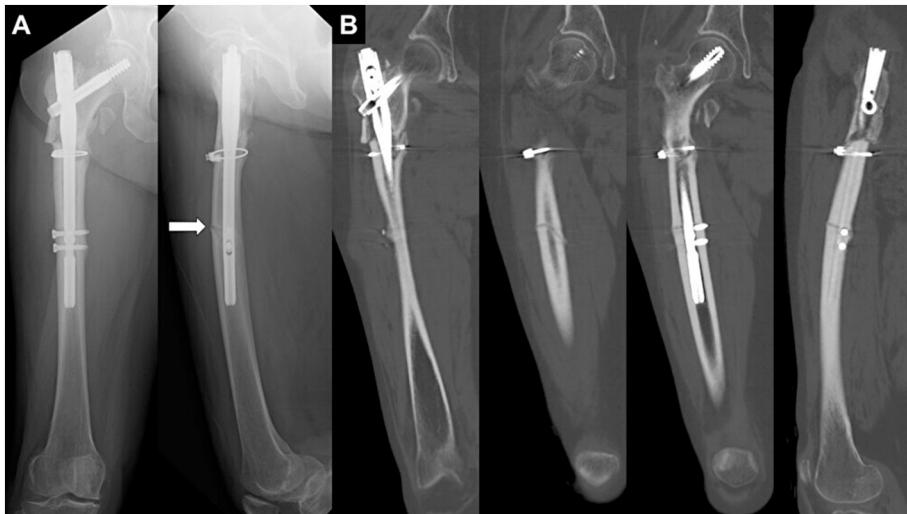


Fig. 3. Plain radiographs and computed tomography (CT) images of the right femur 1 year postoperatively. (A) An anteroposterior view of the radiograph shows no abnormal findings. However, a lateral view of the radiograph shows an incomplete fracture line at the anterior part of the distal screw-insertion site (arrow). (B) CT images show a subtrochanteric femoral nonunion and an incomplete fracture at the anterior part of the distal screw-insertion site.

implant subtrochanteric femoral fractures occur when proximal transverse screws are inserted around the subtrochanteric region. In femurs with a little or no bowing, the maximum tensile force is applied to the subtrochanteric region, and if the transverse-directed screws are inserted around the subtrochanteric region, excessive stress may be concentrated on the subtrochanteric region, resulting in fractures [2,5,6]. In the present case, the varus deformity of the proximal fragment gradually progressed, and the distal part of the mid-length nail was laterally displaced, which indicated an excessive stress concentration around the distal screw-insertion site, resulting in the diaphyseal fracture.

The patient complained of pain, and a contralateral atypical diaphyseal femoral fracture had occurred, which allowed early detection of an atypical diaphyseal femoral fracture after intramedullary nailing for an atypical subtrochanteric femoral fracture. In secondary atypical femoral fractures, radiolucent fracture lines are often masked by preexisting implants used for prior fracture fixation. Especially in cases of intramedullary nailing, fracture lines that run transversely are hidden by screws and are difficult to detect on the anteroposterior view of radiographs because fractures most often occur around the screw-insertion site. To detect fractures before the bone breaks completely, Kim et al. [6] recommended the use of the lateral view of radiographs or bone scintigraphy.

The treatment of secondary atypical femoral fractures depends on the fracture location, prior implants used for femoral fixation,

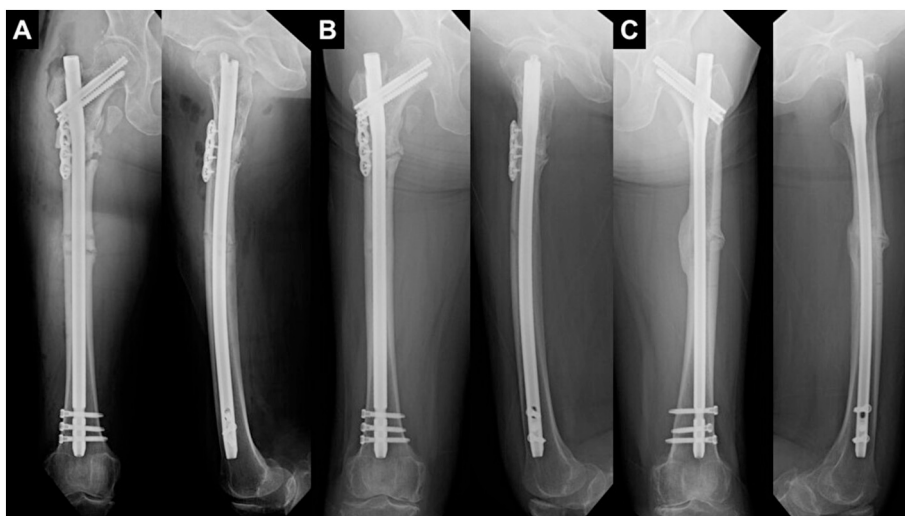


Fig. 4. Radiographs after the revision surgery and at the final follow-up. (A) A long-length nail is inserted after realignment and provisional plate fixation of the subtrochanteric lesion. (B) One and a half years after the revision surgery, bony union of the diaphyseal lesion is obtained, but the subtrochanteric lesion is not healed. (C) Two years after the surgery, bony union of the diaphyseal lesion is obtained.

and whether the fractures are complete. For secondary atypical subtrochanteric femoral fractures, the proximal transverse screws were removed and replaced with screws directed toward the femoral head, or only the subtrochanteric screw was removed to avoid stress concentration in patients who originally had screws inserted into the femoral head and subtrochanteric region [3–6]. For secondary atypical diaphyseal femoral fractures, which reportedly occur only after plate fixation, plate removal and refixation using long-length intramedullary nails were performed in all but one case [6]. In the revision surgery in the present case, a long-length nail was inserted after realignment and provisional plate fixation to achieve bony union of the subtrochanteric and diaphyseal lesions. In addition, to prevent varus displacement of the proximal femoral fragment during nail insertion, the nail entry was reopened more medially [8].

The appropriate length of the intramedullary nail for atypical subtrochanteric femoral fracture fixation is controversial. Kim et al. [9] compared the clinical outcomes of partial- and full-length nails used for atypical subtrochanteric femoral fractures and concluded that full-length nails are not usually required. According to their report, no ipsilateral secondary atypical femoral fractures were found during the follow-up period; however, the mean follow-up period was a short 1.9 years. Although a secondary atypical diaphyseal femoral fracture was found 1.5 years after the initial surgery in the present case, secondary atypical femoral fractures have reportedly occurred an average of 6.6 years after the initial surgery [6], and secondary atypical femoral fractures may occur with longer follow-up periods, especially in the partial-length nail group. For the treatment of atypical subtrochanteric femoral fractures, long-length nails and proximal screw insertion directed toward the femoral head are preferred to prevent secondary atypical diaphyseal femoral fractures.

Conclusion

Secondary atypical diaphyseal femoral fractures after mid-length intramedullary nailing for atypical subtrochanteric femoral fractures can occur because of the stress concentration around the distal screw-insertion site. For atypical subtrochanteric femoral fractures, the use of long-length nails and proximal screw insertion directed toward the femoral head may be important to prevent secondary atypical diaphyseal femoral fractures.

Ethical approval

Ethical approval is not a requirement at our institution for reporting individual cases or case series.

Consent

Written informed consent was obtained from the patient for the publication of this case report and accompanying images. A copy of the written consent form is available for review by the journal's editor-in-chief upon request.

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Ethical approval

The Ethics Committee of the Misawa City Hospital, to which the author belongs, does not require ethics review for case reports of medical procedures performed within the normal scope of care if written consent is obtained from the individual patient.

Guarantor

The guarantor for the present case report is Shuya Nohmi.

CRedit authorship contribution statement

Shuya Nohmi: Writing – original draft. **Masakazu Kogawa:** Data curation. **Taro Ogawa:** Writing – review & editing.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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