Acta Med. Okayama, 2022
Vol. 76, No. 3, pp. 317–321

Acta Medica Okayama
http://escholarship.lib.okayama-u.ac.jp/amo/

Case Report

Hip Fractures after Intramedullary Nailing Fixation for Atypical Femoral Fractures: Three Cases

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Secondary hip fractures (SHFs) rarely occur after intramedullary nailing (IMN) fixation without femoral neck fixation for atypical femoral fractures (AFFs). We report three cases of older Japanese women who sustained SHFs presumably caused by osteoporosis and peri-implant stress concentration around the femoral neck after undergoing IMN without femoral neck fixation for AFF. All cases were fixed with malalignment. In AFF patients, postoperative changes due to postoperative femoral bone malalignment may affect the peri-implant mechanical environment around the femoral neck, which can result in insufficiency fractures. At the first AFF surgery, we recommend femoral neck fixation after adequate reduction is achieved.

Key words: atypical femoral fracture, bone malalignment, intramedullary nail, femoral neck fracture, hip fracture

Atypical femoral fractures (AFFs) are associated with unique abnormalities in bone metabolism and morphology, and their treatment is challenging [1-3]. AFFs occur in the subtrochanteric or shaft region and are commonly treated with intramedullary nailing (IMN) or cephalomedullary nailing (CMN) [4].

Secondary proximal peri-implant hip fractures rarely occur after reconstruction screw configuration fixation for femoral shaft fractures, including AFFs [5-7]. It was reported that secondary hip fractures (SHFs) occur in 5.4% of patients treated with IMN without femoral neck fixation for femoral shaft fractures, and 6.9% for AFFs at the shaft region [7]. Femoral neck bone density and strength are lower in mid-shaft AFFs [6]. Knowledge of these fractures is necessary to elucidate the pathogenesis and provide adequate management. No studies have detailed the course of treatment from the time point of AFF diagnoses.

We present three cases of older Japanese women who sustained SHFs presumably caused by osteoporosis and peri-implant stress concentration around the femoral neck, after they had undergone IMN for an AFF. Herein, we focus on the malunion following IMN fixation.

Case Reports

Table 1 presents the characteristics of the three cases. All patients received long-term alendronate treatment for osteoporosis and could walk independently, without aid, before their injuries. The initial surgical approach

Received August 10, 2021; accepted December 14, 2021.
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Conflict of Interest Disclosures: No potential conflict of interest relevant to this article was reported.

Table 1 Brief summary of patient characteristics

Case number	Sex	Height (cm), Weight (kg)	T-score (± SD)	After IMN fixation for AFF (the first fracture)				Secondary hip fracture				
				Malunion	NSA at fracture side (°)	Femoral neck anteversion at fracture side (°)	Femoral neck anteversion at non-fracture side (°)	Pre-injury walking ability	Injury type	Age (years)	Interval from the first fracture (months)	Fracture type
1	Female	130, 37	NA	Valgus	121	26	35	With walker	No fall	84	8	Femoral neck fracture
2	Female	146, 57	Lumbar −2.2, forearm −2.6	Femoral neck anteversion	139	18	−10	Independent	No fall	73	95	Femoral neck fracture
3	Female	148, 38	NA	Valgus, femoral neck anteversion	120	16	−2	Independent	Fall	92	10	Trochanteric fracture

IMN, intramedullary nailing; AFF, atypical femoral fracture; NSA, neck shaft angle; NA, not available; SD, standard deviation.
Note: Minus femoral neck anteversion means femoral neck retroversion.

involved the use of IMN without femoral neck fixation to treat the AFF, and bone union was achieved after the initial surgery.

Case 1. An 83-year-old woman presented with left thigh pain after falling from a standing position. Her medical history included osteoporosis and hypertension. Laboratory blood test results were within normal ranges. Radiographs showed a left femoral shaft fracture, *i.e.*, a complete non-comminuted transverse fracture with localized periosteal thickening of the lateral cortex (Fig. 1A). The contralateral right femoral bone had lateral bowing and localized periosteal thickening of the lateral cortex. This case fulfilled all five major features for an AFF [2]. Based on these findings, we diagnosed an AFF and fixed the fracture with IMN without femoral neck fixation; however, >5° valgus displacement (6°) remained (Fig. 1B) [8]. Bone union was achieved at 7 months postoperatively, and the patient was able to walk with a walking frame.

One month after the bone union, she presented with left hip pain without falling. Radiographs showed a displaced femoral neck fracture (Fig. 1C). The femoral

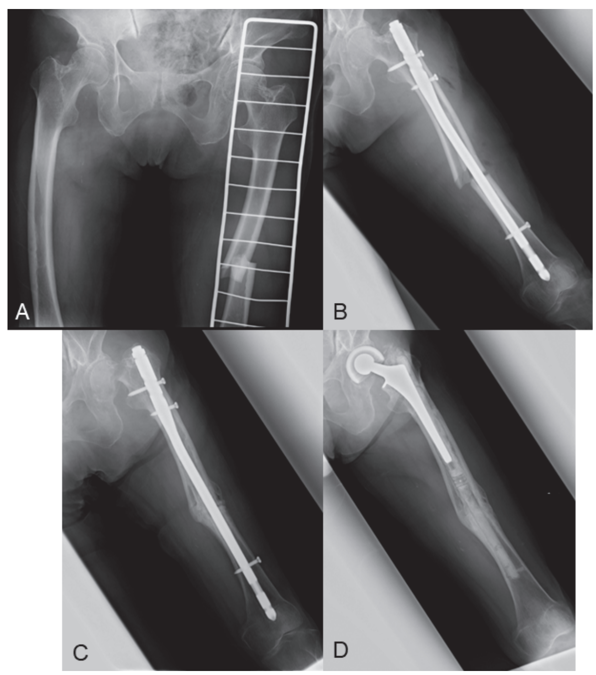

Fig. 1 Case 1. **A**, Radiograph showing the left atypical femoral fracture (AFF). The right femoral bone had lateral bowing and localized periosteal thickening of the lateral cortex; **B**, Postoperative radiograph revealing valgus displacement; **C**, The displaced femoral neck fracture; **D**, Cemented hemiarthroplasty after the nailing's removal.

curvature was 6° valgus and 7° posterior bowing according to the Sasaki method [9]. We removed the IMN and performed a cemented hemiarthroplasty (Fig. 1D). Twelve months postoperatively, the patient was able to walk independently.

Case 2. A 65-year-old woman presented with right thigh pain after falling. Radiographs showed a right AFF in the proximal-shaft region (Fig. 2A). The contralateral left femoral bone had a slight lateral bowing and localized periosteal thickening of the lateral cortex. The left femoral neck retroversion measured 10° (rotation difference > 15° compared to the contralateral side) on computed tomography (CT) [10]. Femoral neck anteversion was evaluated by the angle between the femoral neck axis and the line of femoral condyles at the level of the knee using the CT axial plane. The left femur underwent IMN prophylactic fixation (Fig. 3A). We fixed the right fracture with IMN without femoral neck fixation (Fig. 2B). The malrotation with 18° femoral neck anteversion remained (Fig. 3B). The bone union of the right AFF was achieved at 8 months postoperatively.

At age 73 years, the patient presented with right hip pain without falling. Magnetic resonance imaging showed a non-displaced femoral neck fracture with bone marrow edema (Fig. 2C, D). The T-score at the lumbar spine was −2.2. We exchanged the IMN and fixed the femoral neck with two reconstruction screws and one cannulated cancellous screw from the outside nail (Fig. 2E). Three months postoperatively, the patient was able to walk independently, and bone union was achieved.

Case 3. A 90-year-old woman presented with right thigh pain after falling. Radiographs showed a right AFF in the mid-shaft region (Fig. 4A). We fixed the fracture with retrograde IMN; however, the slight (< 5°) valgus displacement of 3° remained (Fig. 4B) [8].

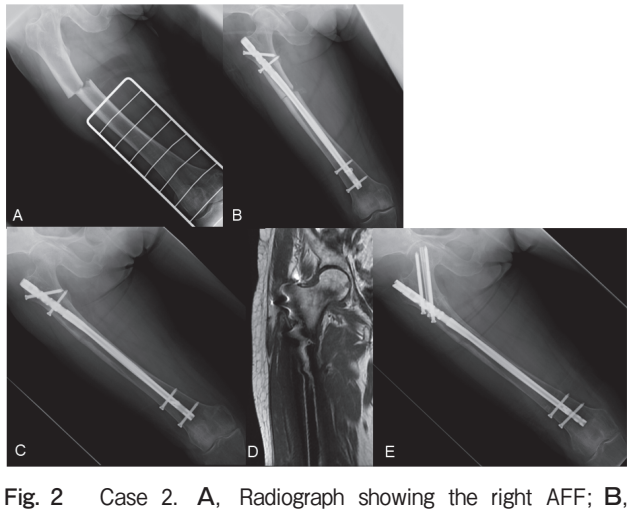

Fig. 2 Case 2. **A**, Radiograph showing the right AFF; **B**, Postoperative radiograph showing the intramedullary nail (IMN) fixation; **C**, The non-displaced femoral neck fracture; **D**, Low signal intensity on T1-weighted images showing bone marrow edema around the femoral neck; **E**, Exchange of the intramedullary nails and screw fixation.

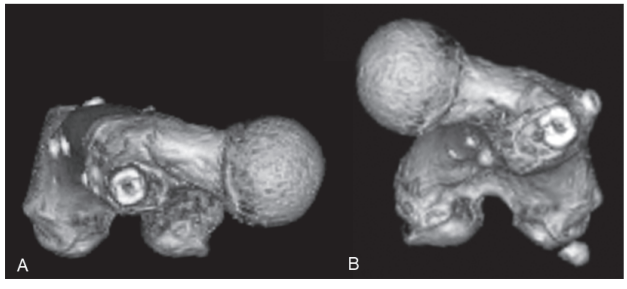

Fig. 3 Case 2. **A**, Postoperative 3D CT of the left femur showing the femoral neck retroversion, which was the original rotation; **B**, Postoperative 3D CT of the right femur showing the femoral neck anteversion, caused by malreduction on rotation.

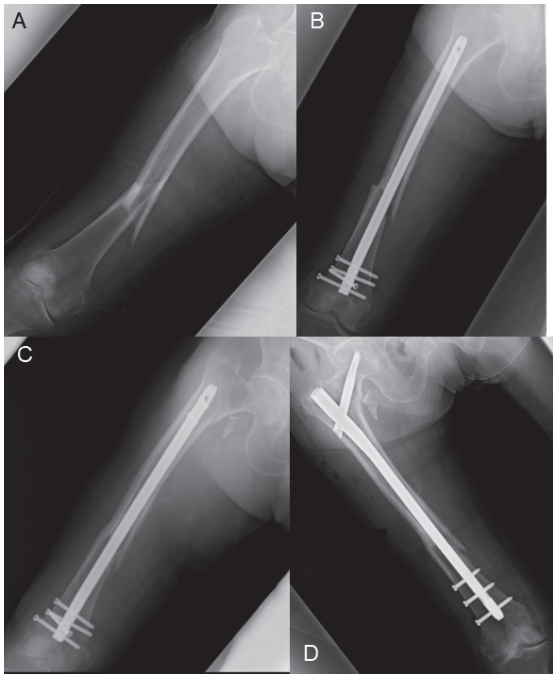

Fig. 4 Case 3. **A**, Radiograph shows right atypical femoral fracture; **B**, Postoperative radiograph shows slight valgus displacement and malrotation after retrograde nailing; **C**, Displaced trochanteric fracture; **D**, Cephalomedullary nail fixation after removing the retrograde nails.

In addition, the malrotation with 16° femoral neck anteversion using CT measurement remained, considering that the left femoral neck retroversion was 2° (rotation difference > 15° compared to the contralateral side) [10]. Bone union was achieved at 3 months postoperatively.

At age 92 years, the patient presented with right hip pain after falling. Radiographs showed an unstable trochanteric fracture, which was classified as AO/OTA 31A2.2 [11] (Fig. 4C). We removed the IMN and fixed the fracture with a long CMN (Fig. 4D). Four months postoperatively, she could walk using a walker, and bone union was achieved.

Discussion

This is the first case series to describe the detailed management course for patients with SHFs associated with malunion following IMN for an AFF. In patients with an AFF, postoperative changes due to postoperative femoral bone malalignment might affect the peri-implant mechanical environment around the femoral neck during loading, which could result in insufficiency fractures.

One potential cause of SHFs after the use of IMN for AFFs is osteoporosis, similar to the risk of an SHF after the use of IMN for typical osteoporotic femoral fractures [12]. We conducted a literature review and identified a few malunion cases (Table 2) [5, 6]. In addition to the factors reported in the previous studies, malrotation and short stature may be relevant (Table 2). Femoral neck malrotation to the non-fractured side is notable because there are no reports about femoral retroversion of AFFs. Considering AFF and postoperative IMN, two other possible rare fracture types are atypical femoral neck fracture and atypical peri-implant fracture [13, 14].

The three present cases might represent examples of combined systemic bone metabolism disorders and repetitive mechanical loading stresses [15]; *i.e.*, the biological cause is osteoporosis and AFF. The mechanical cause is peri-implant and malalignment, wherein the eccentric force might be concentrated in the altered mechanical environment.

There is no consensus regarding the initial nail type selection for an AFF in the shaft region [7]. To the best of our knowledge, there is no published mechanical comparison study comparing the outcomes of the use of

Table 2 Literature review of cases of secondary hip fracture following IMN fixation for an AFF

Authors	Secondary hip fracture cases, n	Sex (female/male)	Malunion following IMN fixation for AFF	Secondary hip fracture					
				Age, years (range)	Injury type	Fracture type (neck fracture/trochanteric fracture)	Interval from the first fracture, mean (range)	Treatment	Reoperation
Schilcher (2000) [5]	2	2/0	1 slight valgus deformity	85 (78–92)	1 fall, 1 NA	1/1	2 years*	1 BHA, 1 sliding nail	1 (failure: sliding nail case)
Tano et al. (2019) [6]	1	1/0	1 slight valgus deformity	83	1 fall	1/0	10 years	Exchange nailing	0
Our study (2021)	3	3/0	1 anterior rotation deformity, 1 valgus deformity, 1 anterior rotation and valgus deformity	83 (73–92)	1 fall, 2 no fall	2/1	3.1 years (0.7–7.9)	1 BHA, 2 Exchange nailing	0

IMN, intramedullary nailing; AFF, atypical femoral fracture; BHA, bipolar hip arthroplasty; NA, not available, *data of only one case is available.

a reconstruction screw and that of the standard proximal interlocking screw in IMN of femoral shaft fractures. Comparative clinical studies of femoral shaft fractures have revealed similar complications [16]. For AFFs, proximal fixation with reconstruction screws would not be inferior to that with standard proximal interlocking screws, and therefore, whole proximal femur bone fixation may be an option for bone healing by providing adequate initial fixation for an AFF and the prevention of a secondary hip fracture in the future.

Our three patients' cases offer three clinical implications. First, a preoperative CT assessment of the whole femur on the non-fractured side should be performed to determine the inherent femoral rotation, because the femoral neck is occasionally in retroversion in AFF cases. Second, surgeons should perform adequate reduction without malalignment. Finally, secondary hip fractures might be prevented using CMN or IMN with reconstruction screws for the first AFF fixation.

In conclusion, this is the first case series of SHFs associated with malunion following IMN for an AFF. At the first surgery for an AFF, we recommend femoral neck fixation using CMN or IMN with reconstruction screws.

Acknowledgments. The English editing fee was supported by the Systematic Review Workshop Peer Support Group (a not-for-profit organization). The funders played no role in the design of the study and collection, analysis, and interpretation of data, or in writing the manuscript.

This work was supported by the Systematic Review Workshop Peer Support Group (SRWS-PSG).

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