TRAUMA SURGERY



A rare case of bilateral proximal femoral insufficiency fractures after interlocking IM nailing for bilateral atypical femoral shaft fractures

Young Sung $\rm Kim^1\cdot Phil Hyun \ Chung^1\cdot Suk \ Kang^1\cdot Jong \ Pil \ Kim^{1,2}\cdot Ho \ Min \ Lee^1\cdot Han \ Sol \ Moon^1$

Received: 13 March 2015/Published online: 14 June 2015 © Springer-Verlag Berlin Heidelberg 2015

Abstract Atypical femoral fractures have unique radiologic and clinical feature. Most commonly used fixation method for atypical femoral fracture is interlocking intramedullary (IM) nailing. The aim of this paper is to document a rare case of a 76-year-old female who sustained bilateral proximal femoral insufficiency fractures after conventional interlocking IM nailing for bilateral atypical femoral shaft fractures without taking history of bisphosphonates. We recommend the routine use of full-length reconstruction or cephalomedullary-type interlocking IM nail for atypical femoral fractures.

Keywords Femoral shaft · Atypical fracture · Insufficiency fracture · Interlocking IM nailing

Introduction

Atypical femoral fractures are characterized by transverse fracture line, medial spike, localized periosteal, or endosteal thickening at the fracture site, minimal, or no comminuted fracture on radiology and have the clinical features of the prodromal symptom, minor trauma history [1–3]. In the recent decade, atypical femoral fractures have been reported in patients taking bisphosphonates (BPs) [3–6],

☑ Jong Pil Kim kjpil@dongguk.ac.kr but they also occur in patients with no exposure to this medication [7, 8], which may be associated with severely suppressed bone turnover. Lower extremity geometry and Asian ethnicity may contribute to the risk of atypical femur fractures [8–10]. Standard treatment regimen is still not established, but most commonly used fixation method is interlocking intramedullary (IM) nailing [11–13].

Here, the authors present a rare case of a 76-year-old female who sustained bilateral proximal femoral insufficiency fractures after conventional interlocking IM nailing for bilateral atypical femoral shaft fractures without taking history of BPs.

Case report

A 76-year-old woman presented to emergency room with left thigh pain and deformity after slip down at toilet. Physical examination revealed swelling and tenderness on her mid thigh and her sensory, motor, and circulation of lower extremity were normal. Simple radiographs showed a transverse, noncomminuted complete fracture with medial spike in the area of the thickened cortex of the femoral diaphysis at 8 cm below the lesser trochanter (Fig. 1). T-score of her lumbar vertebra on DEXA bone mineral density examination was -3.6 but she denied any medical history including treatment of osteoporosis.

The atypical femoral fracture was stabilized by antegrade interlocking IM nailing [cannulated femoral nail (CFN), Ø 13 × 340 mm, AO Synthes[®], Swiss] (Fig. 2). We applied two transverse interlocking screws for each proximal and distal locking area. The patient recovered without any complication and fracture was united at postoperative 3 months. No osteoporosis medication was administrated.

¹ Department of Orthopedic Surgery, College of Medicine, Dongguk University, Gyeongju, Korea

² Department of Orthopaedic Surgery, Dongguk University Gyeongju Hospital, 87 Dongdae-ro, Gyeongju 780-350, Korea



Fig. 1 Initial left femur radiographs of 76-year-old female showed a transverse, noncomminuted complete fracture with medial spike in the area of the thickened cortex of the femoral diaphysis at 8 cm below the lesser trochanter

Three years later, the patient returned with complaints of mild left proximal thigh pain and discomfort. Simple radiographs showed complete fracture union but radiolucent line with cortical hypertrophy at the anterior cortex of femur near distal one of the proximal locking screws (Fig. 3). Diagnosis of stress insufficiency fracture was made, parathyroid hormone therapy was recommended but she declined for personal reasons. Conservative treatment including reducing activity and analgesics was done.

One month later, she presented to emergency room again with right thigh pain and deformity after slip down on floor. Simple radiographs showed a transverse, non-comminuted complete fracture with medial spike in the area of the thickened cortex of the femur midshaft (Fig. 4). This atypical femoral fracture was stabilized by antegrade interlocking IM nailing (CFN, Ø 13 × 340 mm, AO Synthes[®], Swiss) (Fig. 5). Intraoperatively, fracture comminution was happened during inserting the IM nail, we applied two transverse interlocking screws for distal locking, one transverse and one oblique interlocking screw for proximal locking. The fracture was united at postoperative 3 months. No osteoporosis medication was administrated.

Two years later, she returned with complaints of both proximal thigh pain and discomfort. New radiograph showed completely united both femoral shaft fracture, but



Fig. 2 The fracture was stabilized by antegrade interlocking IM nailing with CFN



Fig. 3 Three years follow-up radiographs showed complete fracture union, but radiolucent line with cortical hypertrophy at the anterior cortex of femur near distal one of the proximal locking screws (*arrow*)



Fig. 4 Simple radiographs of right femur after slip down showed a transverse, noncomminuted complete fracture with medial spike in the area of the thickened cortex of the femur midshaft



Fig. 5 The right femur atypical fracture was fixed with interlocking IM nail. The fracture comminution was happened during inserting the IM nail

radiolucent line with cortical hypertrophy at the anterior cortex near distal one of the proximal locking screws of the both femur (Fig. 6). Diagnosis of bilateral proximal femoral stress insufficiency fractures was made. The symptoms were mild, and conservative treatment was done. Three years later at final follow-up postoperative 8 years for left, 5 years for right, she complained intermittent mild pain but she could doing usual daily activity. Radiograph showed no interval change at left side, but radiolucent line and cortical hypertrophy were more defined at anterolateral cortex near distal one of proximal locking screws at right side. The patient did not want to further evaluations or treatments.

Discussion

Atypical femoral fractures considered as the insufficiency stress fractures that progress over time [1]. The insufficiency stress fracture occurs when normal stress is placed on a bone with deficient elastic resistance [14]. Several case reports and case series have described unique radiologic features of atypical femoral fractures with prolonged BP use including a transverse, noncomminuted fracture at the subtrochanteric or femoral shaft area with a medial cortical spike, the exact cause of these fractures is unknown, and likely multifactorial. In many aspects, these fractures have different characters from typical osteoporotic fractures of femur including the injury mechanism, location, fracture configuration, and prodromal symptoms. In 2010, the American Society of Bone and Meneral Research (ASBMR) task force published the first report on atypical femoral fractures defined major and minor features for diagnosis [15]. But, original ASBMR criteria still had some controversial, they revised the case definition in 2014 to highlight radiologic features that distinguish atypical femoral fractures from ordinary osteoporotic diaphyseal femoral fractures [1]. The major features of revised case definition of atypical femoral fractures being association with minimal or no trauma, substantially transverse orientation at the lateral cortex, medial spike when the fracture is complete, non or minimal comminution, localized periosteal or endosteal thickening of the lateral cortex at the fracture site, and minor features being association with cortical thickening, prodromal pain, bilaterality, delayed healing, and removed drug exposures as one of the association from the minor features [1]. Association with lower extremity biomechanical change like a severe curved femur [8, 9, 16], tibio-femoral misalignment [17], or varus hip geometry [18] with atypical femoral fractures were reported. The lateral aspect of the femur was subjected to tensile strains during a variety of physical activities and peak tensile strains during most of walking stance phase [19].

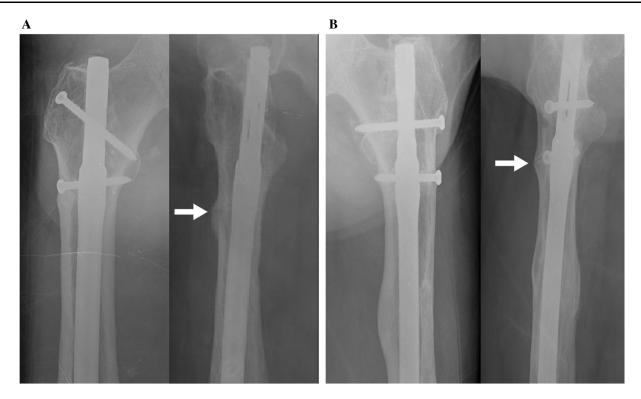


Fig. 6 Follow-up both femur radiographs showed completely united both femoral shaft fracture, but radiolucent line with cortical hypertrophy at the anterior cortex near distal one of the proximal locking screws of the both femur (*arrow*). **a** Right. **b** Left

Among Asian population, stress fracture of the bowed femoral shaft is considered another cause of atypical femoral fracture in elderly person [8-10]. Oh et al. [9] reported a significant tensile stress concentration on the anterolateral surface of femur due to bowing deformity is pathogenic mechanism of atypical femoral fracture through mechanical analysis by CT-based finite element method. Cho and Suh [16] report a rare case of atypical femoral diaphyseal fracture occurring below a previously fixed compression hip screw fixation in laterally bowered femur. Saleh et al. [20] reported the association between typical locations for atypical femoral fractures and tensile strain. Unnanuntana et al. [21] reported long-term alendronate did not appear to cause thickened femoral cortex compared to untreated group and Giusti et al. [22] reported the cortical thickness showed no significant difference between the atypical fracture group and ordinary fracture group. These findings might suggest the lower correlation of BP and atypical femoral fracture. Furthermore, decreased cortical elasticity or severity of osteoporosis itself may be a risk factor for atypical femoral fracture [10]. In our case, the patient had all major and minor features of ASBMR criteria of atypical femoral fracture except a prodromal symptom. But she had anterior bowing deformity of the both femur and osteoporosis without history of BP intake.

The treatment and its effectiveness have not been adequately reported. The recommendation of the ASBMR

task force for medical treatment of atypical femoral fractures are discontinuation of BPs, adequate supply of calcium and vitamin D, and consideration of teriparatide for those who appear not to heal with conservative therapy [1]. For complete atypical femoral fractures, surgical fixation is needed [1, 23, 24]. The majority of incomplete atypical femoral fractures progress to complete fracture, prophylactic surgical treatment should be considered [12, 25, 26].

Even though a higher failure and a high complication rate of IM nailing in BP-associated femur fractures were reported [23, 24], interlocking IM nailing is more preferable fixation method than fixation with plate and screws in atypical femoral fracture [1, 11–13, 26]. A fracture treated with IM nail heals by endochondral repair, whereas a plateand-screw construct generally precludes the endochondral repair process [11]. Oh et al. [12] reported 100 percent union rate after prophylactic nailing for incomplete atypical femoral fractures. Some studies on plate use for atypical femur fractures have reported high failure rate [15, 24, 26]. There is no common consensus that which type of IM nail should be used for atypical femoral fractures. Most authors used standard interlocking nail in mid and distal shaft fractures [12] and cephalomedullary or reconstruction-type nail for proximal subtrochanteric fractures [12, 26]. Oh et al. [8] preferred to use a high-bowing IM nail, such as the Sirus nail (Zimmer GmbH, Winterthur,

Switzerland) for internal fixation for stress fractures associated with a bowed femoral shaft.

The most frequently occurred complication of IM nailing of atypical fractures was intraoperative femoral shaft comminution during nail insertion [24] and nonunion [23, 26]. Fang et al. [27] reported a case of secondary displacement of fracture despite prophylactic antegrade IM nail fixation for atypical undisplaced femoral diaphyseal fracture. Oh et al. [8] have experienced 2 cases of hip fractures during follow-up of atypical femoral fracture after interlocking IM nailing and their recent practice is to insert proximal screws toward the femoral head. Bonifacio and Syson [28] reported a case of construct failure in an atypical subtrochanteric femoral fracture treated with IM nailing. The device was an 11 mm diameter simplified universal nail had a transverse break at the proximal static interlocking screw hole.

In presenting case, bilateral proximal femoral stress insufficiency fractures were occurred after conventional interlocking IM nailing for atypical femoral shaft fractures at anterior cortex near distal one of the proximal locking screws of the both femur. Fang et al. [27] reported a unique case of a displaced subtrochanteric stress fracture with bending of the IM nail after standard interlocking IM nailing for atypical distal femoral shaft fracture. The subtrochanteric stress fracture and IM nail breakage were occurred at the distal one of the proximal locking screws as same as in our presenting case. The proximal portion of the femur is subjected to large bending forces, tensile stresses are applied to the lateral cortex by the geographic features of the femur [19]. We thought the location of maximal tensile strain moved to more proximal area after fixation with interlocking IM nail, repeated tensile stress accumulation at this site might be cause of the insufficiency fractures of the osteoporotic bone. These fractures were also transverse nature but occurred at anterior cortex. Although tensile strain may increase in the anterior of the femoral shaft and make a transverse insufficiency fracture in lordotic femur, the fractures in our case might be stress shielding effect of IM nail and two transverse interlocking screws. Sufficient length, diameter, and cephalomedullarytype of IM nail is recommended to reduce the risk of another stress fracture at the area of load concentration. If distal femoral cortex is too thin, angular stable locking system[®] (ASLS[®], Synthes GmbH, Oberdorf, Switzerland) is helpful to enhance the stability of distal locking screw between IM nail and bone cortex [13].

The authors experienced a case of a 76-year-old female who sustained bilateral proximal femoral insufficiency fractures after conventional interlocking IM nailing for bilateral atypical femoral shaft fractures without taking history of BPs. We recommend the routine use of full-length reconstruction or cephalomedullary-type interlocking IM nail for atypical femoral fractures to prevent the proximal femoral insufficiency fracture even when the fracture is located at mid or distal shaft area.

Conflict of interest The authors have no conflicts of interest to declare.

References

- Shane E, Burr D, Abrahamsen B et al (2014) Atypical subtrochanteric and diaphyseal femoral fractures: second report of a task force of the American Society for Bone and Mineral Research. J Bone Miner Res 29:1–23. doi:10.1002/jbmr.1998
- Bronson WH, Kaye ID, Egol KA (2014) Atypical femur fractures: a review. Curr Osteoporos Rep 12:446–453. doi:10.1007/ s11914-014-0239-7
- Im GI, Jeong SH (2015) Pathogenesis, management and prevention of atypicalfemoral fractures. J Bone Metab 22:1–8. doi:10.11005/jbm.2015.22.1.1
- Gedmintas L, Solomon DH, Kim SC (2013) Bisphosphonates and risk of subtrochanteric, femoral shaft, and atypical femur fracture: a systematic review and metaanalysis. J Bone Miner Res 28:1729–1737
- Schilcher J, Koeppen V, Aspenberg P, Michaëlsson K (2015) Risk of atypicalfemoral fracture during and after bisphosphonate use. Acta Orthop 86:100–107. doi:10.3109/17453674.2015. 1004149
- Rifai A, Pourtaheri S, Carbone A, Callaghan JJ, Stadler CM, Record N, Issa K (2015) Bilateral femur fractures associated with short-term bisphosphonate use. Orthopedics 38:e139–e142. doi:10.3928/01477447-20150204-90
- Tan SC, Koh SB, Goh SK, Howe TS (2011) Atypical femoral stress fractures in bisphosphonate-free patients. Osteoporos Int 22:2211–2212. doi:10.1007/s00198-010-1384-1
- Oh Y, Wakabayashi Y, Kurosa Y, Ishizuki M, Okawa A (2014) Stress fracture of the bowed femoral shaft is another cause of atypical femoral fracture in elderly Japanese: a case series. J Orthop Sci 19:579–586. doi:10.1007/s00776-014-0572-9
- Oh Y, Wakabayashi Y, Kurosa Y, Fujita K, Okawa A (2014) Potential pathogenic mechanism for stress fractures of the bowed femoral shaft in the elderly: mechanical analysis by the CT-based finite element method. Injury 45:1764–1771. doi:10.1016/j. injury.2014.08.037
- Sasaki S, Miyakoshi N, Hongo M, Kasukawa Y, Shimada Y (2012) Low-energy diaphyseal femoral fractures associated with bisphosphonate use and severe curved femur: a case series. J Bone Miner Metab 30:561–567. doi:10.1007/s00774-012-0358-0
- Unnanuntana A, Saleh A, Mensah KA, Kleimeyer JP, Lane JM (2013) Atypical femoral fractures: what do we know about them? AAOS exhibit selection. J Bone Jt Surg Am 95(e8):1–13. doi:10. 2106/JBJS.L.00568
- Oh CW, Oh JK, Park KC, Kim JW, Yoon YC (2013) Prophylactic nailing of incomplete atypical femoral fractures. Sci world J 2013:450148. doi:10.1155/2013/450148
- Lee KJ, Kwon YW, Ha YC, Lee YK, Kim BS, Min BW (2013) Surgical treatment of atypical femoral fracture. J Korean Orthop Assoc 48:185–189
- Daffner RH, Pavlov H (1992) Stress fracture current concepts. AJR AM J Roentgentol 159:245–252
- 15. Shane E, Burr D, Ebeling PR et al (2010) Atypical subtrochanteric and diaphyseal femoral fractures: report of a task force of the American Society for Bone and Mineral Research. J Bone Miner Res 25:2267–2294

- Cho HJ, Suh KT (2015) A rare case of a bisphosphonate-related femoral diaphyseal fracture occurring below a previously fixed compression hip screw (CHS) plate. J Orthop Sci 20:559–562. doi:10.10007/s00776-013-0519-6
- Saita Y, Ishijima M, Mogami A et al (2014) The fracture sites of atypical femoral fractures are associated with the weight-bearing lower limb alignment. Bone 66:105–110. doi:10.1016/j.bone. 2014.06.008
- Hagen JE, Miller AN, Ott SM et al (2014) Association of atypical femoral fractures with bisphosphonate use by patients with varus hip geometry. J Bone Jt Surg Am 96:1905–1909. doi:10.2106/ JBJS.N.00075
- Martelli S, Pivonka P, Ebeling PR (2014) Femoral shaft strains during daily activities: implications for atypical femoral fractures. Clin Biomech 29:869–876. doi:10.1016/j.clinbiomech.2014.08. 001
- Saleh A, Hegde VV, Potty AG, Schneider R, Cornell CN, Lane JM (2012) Management strategy for symptomatic bisphosphonate-associated incomplete atypical femoral fractures. HSS J 8:103–110. doi:10.1007/s11420-012-9275-y
- Unnanuntana A, Ashfaq K, Ton QV, Kleimeyer JP, Lane JM (2012) The effect of long-term alendronate treatment on cortical thickness of the proximal femur. Clin Orthop Relat Res 470:291–298
- 22. Giusti A, Hamdy NAT, Dekkers OM, Ramautar SR, Dijkstra S, Papapoulos SE (2011) Atypical fractures and bisphosphonate

therapy: a cohort study of patients with femoral fracture with radiographic adjudication of fracture site and femurs. Bone 48:966-971

- Weil YA, Rivkin G, Safran O, Liebergall M, Foldes AJ (2011) The outcome of surgically treated femur fractures associated with long-term bisphosphonate use. J Trauma 71:186–190. doi:10. 1097/TA.0b013e31821957e3
- Prasarn ML, Ahn J, Helfet DL, Lane JM, Lorich DG (2012) Bisphosphonate-associated femur fractures have high complication rates with operative fixation. Clin Orthop Relat Res 470:2295–2301. doi:10.1007/s11999-012-2412-6
- 25. Ha YC, Cho MR, Park KH, Kim SY, Koo KH (2010) Is surgery necessary for femoral insufficiency fractures after long-term bisphosphonate therapy? Clin Orthop Relat Res 468:3393–3398. doi:10.1007/s11999-010-1583-2
- 26. De Das S, Setiobudi T, Shen L (2010) A rational approach to management of alendronate-related subtrochanteric fractures. J Bone Joint Surg Br 92:679–686
- 27. Fang C, Chau JY, Woo SB, Lau TW, Kwan K, Leung F (2014) Propagation of bisphosphonate-related femoral stress fractures despite femoral nailing: a cautionary tale from 2 cases. Geriatr Orthop Surg Rehabil 5:14–17. doi:10.1177/2151458514522058
- Bonifacio L, Syson P (2014) Construct failure in an atypical femoral fracture treated with intramedullary nailing: a case report. Malays Orthop J 8:82-84. doi:10.5704/MOJ.1403.008