

Sequential Subtrochanteric Femoral Fracture after Atypical Diaphyseal Fracture in a Long-Term Bisphosphonate User: A Case Report

Následná subtrochanterická zlomenina femuru po atypické zlomenině diafýzy u dlouhodobého uživatele bisfosfonátů – kazuistika

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SUMMARY

Recent reports have found a relationship between long-term bisphosphonate therapy and the occurrence of low-energy subtrochanteric or diaphyseal atypical femoral fractures. These fractures usually occur at only one site in the same bone. We report a rare case of a patient with sequential atypical femoral fractures (first, a diaphyseal fracture, and second, an ipsilateral subtrochanteric fracture) after low-energy trauma. We present the clinical and operative findings and discuss how to prevent subsequent atypical femoral fractures. This case indicates that an atypical subtrochanteric femoral fracture can occur after an atypical diaphyseal fracture in a long-term bisphosphonate user. Doctors should be aware of the possibility of a second fracture and explain the risk to the patient.

Key words: bisphosphonate, atypical femoral fracture, low-energy trauma, subtrochanteric and diaphyseal femoral fractures.

This study was supported by a grant from the CNUH-BRI (Biomedical Research Institute of Chonbuk National University Hospital, CNUH-BRI-2012-02-005)

INTRODUCTION

As a group, atypical femoral fractures (AFFs) have strikingly similar clinical and radiological features. They occur in the subtrochanteric or diaphyseal region of the femur and have a simple transverse or oblique pattern, without comminution (4, 10, 12). They occur after minimal trauma; are often preceded by prodromal pain, which may be of several months' duration, and are bilateral in approximately 50% of cases (3, 7). Cortical thickening and beaking are commonly observed, and healing is often delayed (6, 13). Atypical fractures appear to originate in the lateral cortex, where they may show features characteristic of a stress fracture with a periosteal reaction (1).

A few studies have reported the occurrence of an atypical femoral fracture at one site (diaphyseal or subtrochanteric) of the femur after long-term use of bisphosphonate (1, 10); however, no reports have docu-

mented the occurrence of an ipsilateral subtrochanteric fracture after a diaphyseal fracture.

Here, we report on a rare case of a patient with sequential atypical femoral fractures (first, a diaphyseal fracture, and second, an ipsilateral subtrochanteric fracture) after low-energy trauma. The atypical diaphyseal femoral fracture occurred due to minimal trauma (i.e., a falling during walking). After intramedullary nailing for the diaphyseal fracture, an ipsilateral subtrochanteric femoral fracture occurred due to minimal trauma (i.e., a falling during crutch ambulation).

CASE REPORT

A 74-year-old woman presented with right thigh pain after a falling during walking with a crutch. She had a medical history of postmenopausal osteoporosis. In

this patient, menopause occurred at age 50, and she had been on bisphosphonate for 10 years for osteoporosis, which was diagnosed by dual energy X-ray absorptiometry (DEXA).

Initial plain AP lateral radiographs of the right femur showed diffuse cortical thickening and beaking and excessive anterior bowing (Fig. 1A, B). At that time, the surgeon did not inform the patient about the management of an atypical femoral fracture and instead prescribed additional bisphosphonate medication. The patient fell down in the bathroom and incurred a transverse fracture on the distal one-third of the right femoral shaft (Fig. 1C, D). Closed intramedullary nailing with two proximal transverse locking screws (UFN, AO Synthes, Switzerland) was performed using fracture table at a local clinic (Fig. 2A, B). One week later, during rehabilitation with partial weight-bearing crutch ambulation, the patient fell down and experienced severe pain, after which she was transferred to our hospital. Plain AP lateral radiographs of the right femur showed an ipsilateral oblique subtrochanteric fracture with hardware failure (Fig. 2C, D). We diagnosed this as an atypical femoral fracture due to long-term bisphosphonate use on the basis of specific radiological findings (i.e., diffuse cortical thickening and beaking), drug dosage, and fracture site.

Her T-score in DEXA was -4.0 for the lumbar spine and -3.4 for the femoral neck. The levels of biochemical markers (24-hour urine calcium, C-terminal telopeptide, osteocalcin, bone alkaline phosphatase) were within the normal range.

The original intramedullary nail was removed and fixation was performed with a cephalomedullary intramedullary nail (gamma-3 nail, Stryker, USA), (Fig. 2E, F).

ROM exercises, quadriceps setting exercises, and straight leg raising exercises were recommended after surgery immediately. Two weeks after surgery, partial weight-bearing ambulation was allowed. Within 6 weeks of the operation, full weight-bearing ambulation was possible. At 12 weeks after the operation, bony union without displacement of the fracture site was observed on X-ray, and the pain at the fracture region had disappeared completely. At the patient's final follow-up at 1.5 years after the operation, she reported no complaints with regards to activities of daily living, and bony union was observed on plain X-rays (Fig. 2G, H).

DISCUSSION

The medical management of bisphosphonate-induced atypical femoral fractures is not yet well-established. Until evidence-based recommendations are in place, it may be best for physicians to stop prescribing bisphosphonate. Instead, they should encourage calcium and vitamin D supplementation and should follow up with patients using dual energy X-ray absorptiometry and bone turnover marker measurements (5). Furthermore, treatment with teriparatide (recombinant human parathyroid hormone) may help reverse microdamage in patients previously treated with bisphosphonates (2, 5, 11, 14).

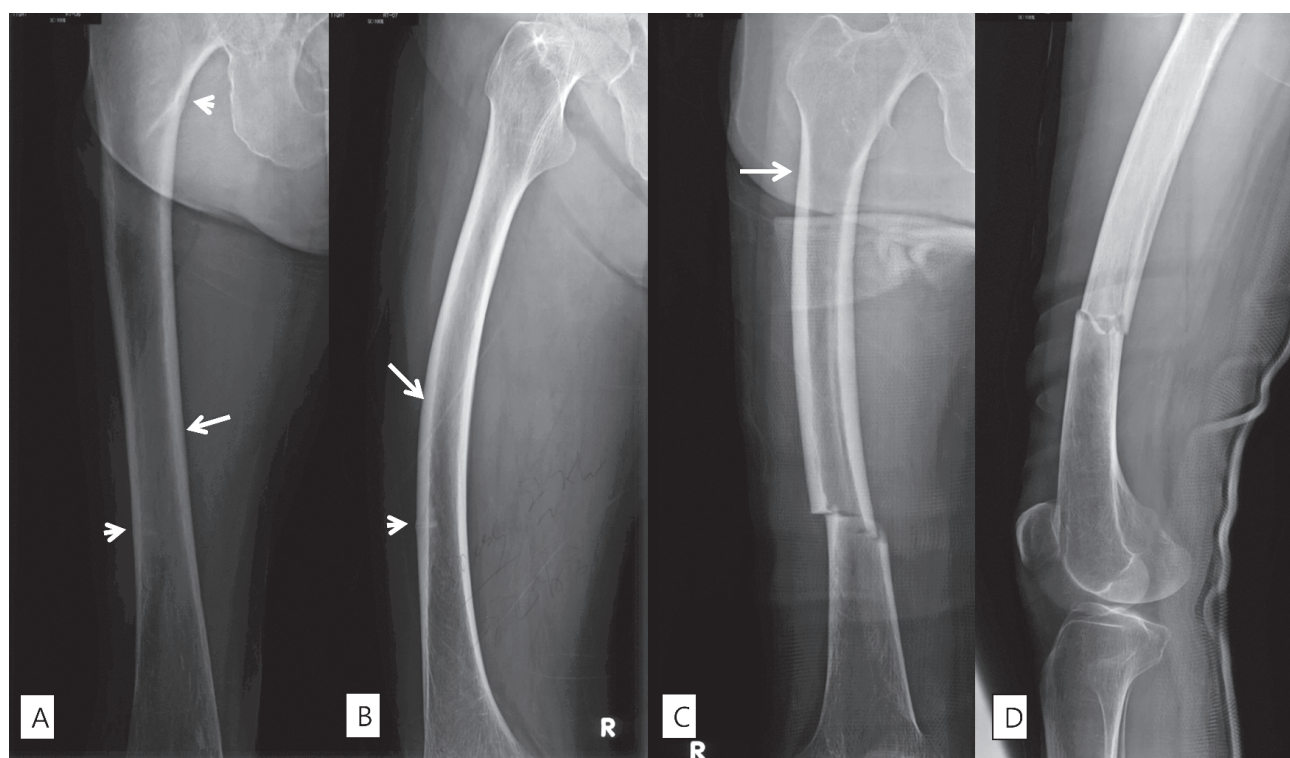


Fig. 1. AP (A) and lateral (B) radiograph of the right femur showing diffuse cortical thickening (white arrow), beaking (arrowhead) and excessive anterior bowing. AP (C) and lateral (D) radiograph of the right femur showing the transverse fracture on the distal one-third of the shaft (first fracture). Note the diffuse cortical thickening from diaphysis to subtrochanter area (white arrow) in Fig. 1C.

Long-term bisphosphonate-induced atypical femoral fractures commonly occur at either the subtrochanteric or diaphyseal regions of the femur (1, 4, 10, 12). To our knowledge, this is the first study to report the serial occurrence of atypical femoral fracture at two different sites, and our results suggest the following: 1) diagnosis or early detection based on X-ray findings suggestive of an atypical femoral fracture (i.e., beak or cortical thickening) is not easy due to implanted material (intramedullary nailing in this case); 2) lateral femoral bowing frequently occurs in older woman, which produces stress concentration on the medial side of the femur and increases fracture occurrence at this site; and 3) intramedullary nailing with transverse locking screw in the frequent site of an atypical femoral fracture such as subtrochanteric area may increase the risk of a subsequent fracture in this area because proximal locking screws are located around the subtrochanteric area (stress concentration site). In our case, the proximal screws in the first surgery were fixed in the subtrochan-

teric area. Therefore, surgeons should be aware of the possibility of a second fracture and think other fixation method like cephalomedullary type.

It is very important for a surgeon to consider the best choice of surgical method, including the instrument type, in the treatment of an atypical femoral fracture because a silent stress fracture due to long-term bisphosphonate use can subsequently occur at another site. When the femur is fractured in the diaphyseal region, the best treatment may be cephalomedullary type (i.e., proximal interlocking screws to the femoral head) instead of intramedullary nailing with transverse locking screws because cephalomedullary treatment can cover a possible fracture in the subtrochanteric area.

Intramedullary nailing is currently accepted as the gold standard in the treatment of diaphyseal femur fractures (2). However, surgeon should consider the nail insertion point move slightly anterior to the junction of the femoral neck and the greater trochanter because there is a mismatch of current intramedullary nails in case

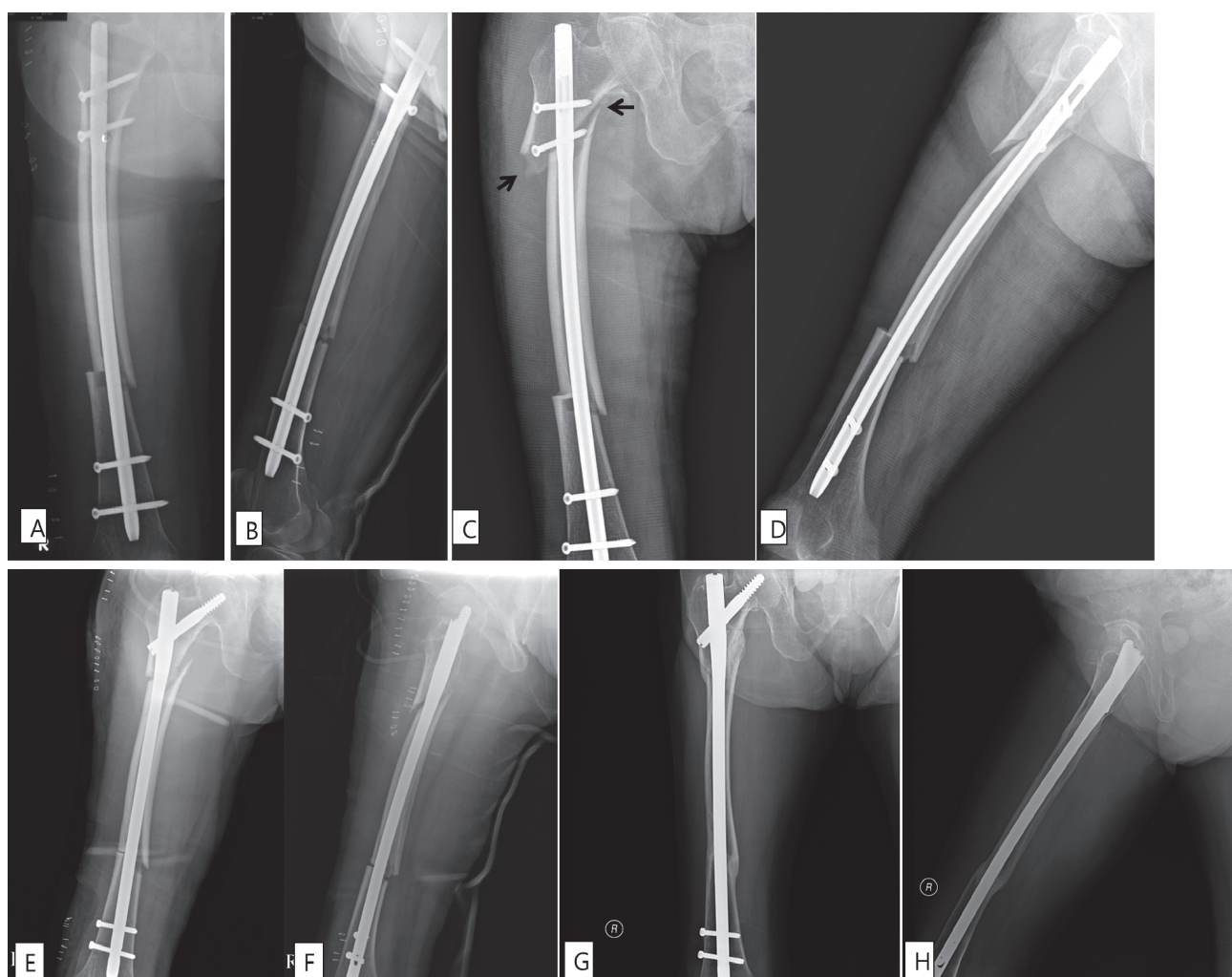


Fig. 2. AP (A) and lateral (B) radiograph of the right femur showing internal fixation with intramedullary nailing with transverse locking screws (UFN, AO Synthes, Swiss). AP (C) and lateral (D) radiograph of the right femur showing the oblique periprosthetic fracture in the subtrochanteric area (secondary fracture, arrows). Postoperative AP (E) and lateral (F) radiograph of the right femur showing internal fixation with a cephalomedullary intramedullary nail (gamma-3 nail, Stryker, USA) and final follow-up AP (G) and lateral (H) radiograph of the right femur showing bony union at the subtrochanteric fracture site.

of excessive anterior bowing of the femur and subsequent complications like anterior placement of nail and anterior cortical penetration can develop (8, 9).

CONCLUSION

In conclusion, an atypical femoral fracture can easily occur not only on the diaphysis but also in the sub-

trochanteric area. Our results suggest that during proximal screwing to the subtrochanteric area during intramedullary nailing for a patient who is a long-term bisphosphonate user, an atypical fracture in the subtrochanteric area can later occur after minimal trauma. The use of cephalomedullary intramedullary nailing can help prevent a subsequent atypical fracture at another site.

References

1. ARMAMENTO-VILLAREAL, R., NAPOLI, N., DIEMER, K., WATKINS, M., CIVITELLI, R., TEITELBAUM, S., NOVACK, D.: Bone turnover in bone biopsies of patients with low-energy cortical fractures receiving bisphosphonates: a case series. *Calcif. Tissue Int.*, 85: 37–44, 2009.
2. BURNEI, C., POPESCU, G. H., BARBU, D., CAPRARU, F.: Intramedullary osteosynthesis versus plate osteosynthesis in subtrochanteric fractures. *J. Med. Life*, 4: 324–329, 2011.
3. CAPECI, C. M., TEJWANI, N. C.: Bilateral low-energy simultaneous or sequential femoral fractures in patients on long-term alendronate therapy. *J. Bone Jt Surg.*, 91-A: 2556–2561, 2009.
4. DELL, R., GREENE, D., OTT, S., SILVERMAN, S., EISEMON, E., FUNAHASHI, T., ADAMS, A.: A retrospective analysis of all atypical femur fractures seen in a large California HMO from the years 2007 to 2009. *ASBMR 2010 Annual Meeting*, Toronto, Canada, 2010, 1201.
5. DOBNIG, H., STEPAN, J. J., BURR, D. B., LI, J., MICHALSKA, D., SIPOS, A., PETTO, H., FAHRLEITNER-PAMMER, A., PAVO, I.: Teriparatide reduces bone microdamage accumulation in postmenopausal women previously treated with alendronate. *J. Bone Miner. Res.*, 24: 1998–2006, 2009.
6. GIUSTI, A., HAMDY, N. A. T., DEKKERS, O. M., RAMAUTAR, S. R., DIJKSTRA, S., PAPAPOULOS, S. E.: Atypical fractures and bisphosphonate therapy: a cohort study of patients with femoral fracture with radiographic adjudication of fracture site and features. *Bone*, 48: 966–971, 2011.
7. GOH, S. K., YANG, K. Y., KOH, J. S., WONG, M. K., CHUA, S. Y., CHUA, D. T., HOWE, T. S.: Subtrochanteric insufficiency fractures in patients on alendronate therapy: a caution. *J. Bone Jt Surg.*, 89-B: 349–353, 2010.
8. HARMA, A., GERMEN, B., KARAKAS, H. M., ELMALI, N., MUHARREM, I.: The comparison of femoral curves and curves of contemporary intramedullary nails. *Surg. Radiol. Anat.*, 27: 502–506, 2005.
9. HARPER, M. C., CARSON, W. L.: Curvature of the femur and the proximal entry point for an intramedullary rod. *Clin. Orthop. Relat. Res.*, 220: 155–161, 1987.
10. ISAACS, J. D., SHIDIK, L., HARRIS, I. A., SZOMOR, Z. L.: Femoral insufficiency fractures associated with prolonged bisphosphonate therapy. *Clin. Orthop. Relat. Res.*, 468: 3384–3392, 2010.
11. JOBKE, B., PFEIFER, M., MINNE, H. W.: Teriparatide following bisphosphonates: initial and long-term effects on microarchitecture and bone remodeling at the human iliac crest. *Connect Tissue Res.*, 50: 46–54, 2009.
12. LENART, B. A., NEVIASER, A. S., LYMAN, S., CHANG, C. C., EDOBOR-OSULA, F., STEELE, B., VAN DEN MEULEN, M. C., LORICH, D. G., LANE, J. M.: Association of low-energy femoral fractures with prolonged bisphosphonate use: a case control study. *Osteoporos Int.*, 20: 1353–1362, 2009.
13. SHANE, E., BURR, D., EBELING, P. R., ABRAHAMSEN, B., ADLER, R. A., BROWN, T. D., CHEUNG, A. M., COSMAN, F., CURTIS, J. R., DELL, R., DEMPSTER, J., EINHORN, G. A., GENANT, H. K., GEUSENS, P., KLAUSHOFFER, K., KOVAL, K., LANE, J. M., MCKIERNAN, F., MCKINNEY, R., NG, A., NIEVES, J., O KIEFE, R., PAPAPOULOS, S., SEN, H. T., VAN DER MEULEN, M. C. H., WEINSTEN, M. S., WHYTE, M.: 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, 2010.
14. SLOAN, A. V., MARTIN, J. R., LI, S., LI, J.: Parathyroid hormone and bisphosphonate have opposite effects on stress fracture repair. *Bone*, 47: 235–240, 2010.

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