



Case report

Bilateral atypical femoral fracture and end-stage arthritis of the hip, treated with total hip arthroplasty

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ABSTRACT

The prolonged use of bisphosphonates has been associated with an increased rate of atypical femoral fracture. A 77-year-old woman with prolonged bisphosphonate use presented to our office with groin pain and end-stage arthritis. She was scheduled for a total hip replacement. Before the surgery and with minimal trauma, the patient then suffered a displaced atypical femoral fracture. She underwent a total hip replacement as a treatment for her fracture and her arthritis. Subsequently, the patient presented with pain in the contralateral thigh with an incomplete atypical femoral fracture. That side was also treated with a total hip arthroplasty. An uncemented stem with open reduction internal fixation and a long cemented stem were used on the complete fracture and incomplete fracture sides, respectively. At a follow-up of 2 years, the patient had no pain and had excellent function demonstrating the short-term success of both cemented and uncemented stems in total hip arthroplasty after atypical femoral fractures.

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Introduction

Since 2005, it has been reported that long-term bisphosphonate (BP) therapy could be linked to atraumatic low-energy femoral fractures [1]. On occasion, these fractures present with no prodrome. It seems that the prolonged use of BPs may be associated with severe suppression of bone turnover, resulting in atypical insufficiency fractures in unusual locations. The majority of these reports have described fractures in the subtrochanteric region of the femur. Postulated contributory mechanisms include increased advanced glycated end products, increased bone matrix mineralization, and the accumulation of microfractures in the zones of maximal tensile loading combined with suppressed remodeling rates [2–6].

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Several case series have discussed the outcomes of the operative management of atypical femoral fractures (AFFs). Weil et al. [7] reviewed the clinical data from 15 patients with 17 AFFs associated with long-term BP use who were treated surgically. The rate of fracture healing after the initial procedure among patients treated with intramedullary nails was 54%, with the remainder of patients requiring reoperation. The authors attributed this high failure rate to qualitative bone defects caused by long-term BP use. Prasarn et al. [8] compared the rate of intraoperative fractures and postoperative plate failures in 25 patients with BP-related femoral shaft fractures with 20 control patients having similar fracture types but without a history of BP use. The BP group had a significantly higher rate of major complications, including intraoperative fracture, implant failure, nonunion, malunion, and periprosthetic fractures. To assess the efficacy of the nonoperative treatment of BP-related AFFs, Banffy et al. [9] retrospectively reviewed 34 patients with 40 BP-related AFFs. The authors concluded that the nonoperative treatment of these BP-related AFFs is not feasible or reliable because most of these lesions progress to complete fractures.

We present the case of a patient who sustained a bilateral AFF subsequent to the prolonged use of BP and was treated with bilateral total hip arthroplasty (THA) using 2 different methods of

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femoral implant fixation. To our knowledge, this is the first case reported with such treatment modalities.

Case history

The patient was informed that the data concerning her case would be submitted for publication, and the patient agreed.

A 77-year-old Hispanic female initially presented to our office with right groin pain and severe radiographic arthritis of both hips. The patient was using a cane for ambulation. Her medical history was significant for osteoporosis, for which the patient was taking alendronate sodium. In addition, the patient had bilateral total knee arthroplasties performed 15 years before the initial office visit. Her activity level according to the University of California, Los Angeles (UCLA) activity-level rating scale was a 3 out of 10 (sometimes participates in mild activities as walking, limited housework, and limited shopping). The patient's Harris Hip Score was 41 (range, 0–100). On the pain visual analog scale, her right hip pain was graded in both intensity and frequency as 9/10. Preoperatively, her vitamin D level was 33.8 ng/mL and her calcium level was 8.8 mg/dL. On the physical examination, her right hip range of motion was the following: flexion: 100°, abduction: 35°, adduction: 30°, internal rotation: 20°, and external rotation: 40°. The Trendelenburg test was positive with a lurch. She had no deficits in motor strength. The pelvis radiograph showed severe osteoarthritis (OA) of both hips, with the right hip worse than the left (Fig. 1). The patient was therefore diagnosed with end-stage OA of both hips. As the right hip was worse than the left hip, a right THA was planned. Before her surgery, the patient was sent for a bone scan to evaluate a bony island seen on her preoperative hip radiographs (Fig. 1, black circle). The bone scan showed focal activity on the lateral aspect of the proximal right femur, distal to the bone island, and on the midshaft of the left femur; findings were consistent with a most likely diagnosis of an impending AFF based on her medical history and the prolonged use of BP (Fig. 2). However, owing to the nonspecific nature of the bone scan results, the patient was instructed to get a magnetic resonance imaging (MRI) scan to further evaluate these areas of increased signal in both femurs.

Before the date of her THA and planned MRI of her right and left femurs, however, the patient presented to the emergency room with right hip pain after sustaining a fall from a standing height. The patient denied any loss of consciousness or vision, chest pain,

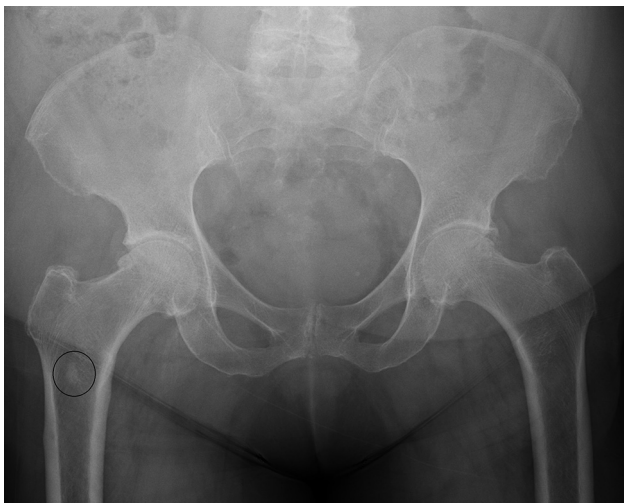


Figure 1. Preoperative anteroposterior pelvis radiograph showing severe bilateral OA of the hips, with the right side worse than the left side.

or dizziness before or after the fall. Her radiograph of the right hip (Fig. 3) at the time of the fall showed severe OA of the right hip and a noncomminuted subtrochanteric femoral fracture with the fracture line beginning at the lateral cortex and continuing medially in a transverse orientation, an AO 32-A3.1 type fracture. This fracture pattern is consistent with an AFF, based on criteria by Shane et al. [1]. Owing to the underlying OA of the right hip, the patient underwent a right THA with open reduction internal fixation of the fracture (Fig. 4). The femoral stem placed was a 140 mm-by-180 mm 70% porous-coated straight revision stem. Fracture treatment was completed using the principles of periprosthetic fracture fixation as described by Shah et al. [10]. The subtrochanteric fracture was further stabilized with 2 fibular strut grafts that were secured in place with Luque wires at 3 levels.

Postoperatively, the patient was placed on our standard anticoagulation protocol for a total of 6 weeks. Her weight-bearing status was toe-touch weight-bearing on the right lower extremity for a total of 6 weeks. She continued to progress well postoperatively in a rehabilitation facility until two and a half weeks after her right THA. At that point, the patient unexpectedly began to complain of severe left hip pain with associated thigh pain. The radiographs (Fig. 5a) and the MRI of her left hip (Fig. 5b and c) showed a thickening of the lateral cortex with a partial cortical disruption of this cortex (white asterisks on Fig. 5b and c) which were interpreted as impending AFFs.

Preoperatively, her activity level (based on the UCLA activity-level rating scale) was rated as a 3 out of 10 (sometimes participates in mild activities as walking, limited housework, and limited shopping). The patient's Harris Hip Score was 41. On the pain visual analog scale, her pain was graded 6.5/10 for intensity and 7/10 for frequency. On the physical examination, her left hip range of motion was the following: flexion: 90°, abduction: 30°, adduction: 25°, internal rotation: 20°, and external rotation: 30°. The patient had no deficits in motor strength.

Because of the underlying and known severe OA of the hip as well as the impending AFF, the patient underwent a cemented left THA (Fig. 6). To allow for adequate fixation of the impending midshaft fracture, a 210-mm cemented revision stem was placed to function as an intramedullary device, as described in the principles of impending AFF fixation by Borrelli et al. [11]. Postoperatively, the patient was restarted on the standard anticoagulation protocol for a total of 6 weeks. In the immediate postoperative period, she was bearing full weight on the left lower extremity. The patient continued to do well postoperatively, and she recovered uneventfully.

At the 1-year follow-up visit, her UCLA activity level was graded as a 6 out of 10 (regularly participates in moderate activities). The patient's Harris Hip Score was 89. On the pain visual analog scale, her pain was graded 0/10 for both intensity and frequency. On the physical examination, her right hip range of motion was as follows: flexion: 95°, abduction: 30°, adduction: 25°, internal rotation: 20°, and external rotation: 45°. Her left hip range of motion was the following: flexion: 90°, abduction: 30°, adduction: 20°, internal rotation: 10°, and external rotation: 60°. She had no deficits in motor strength. The Trendelenburg test was normal. The 1-year follow-up radiographs showed excellent positioning of all components with evidence of healing along the right femur (Fig. 7). Functionally, the patient had minimal difficulty ambulating.

At the latest follow-up visit (2 years), she continues doing well. She has been ambulating without difficulty and is able to get around within the community with minimal difficulty. Her hip range of motion remains excellent, and at this most recent visit, there are no radiographic signs of loosening or instability in both hip arthroplasties (Fig. 8a, b, and c).

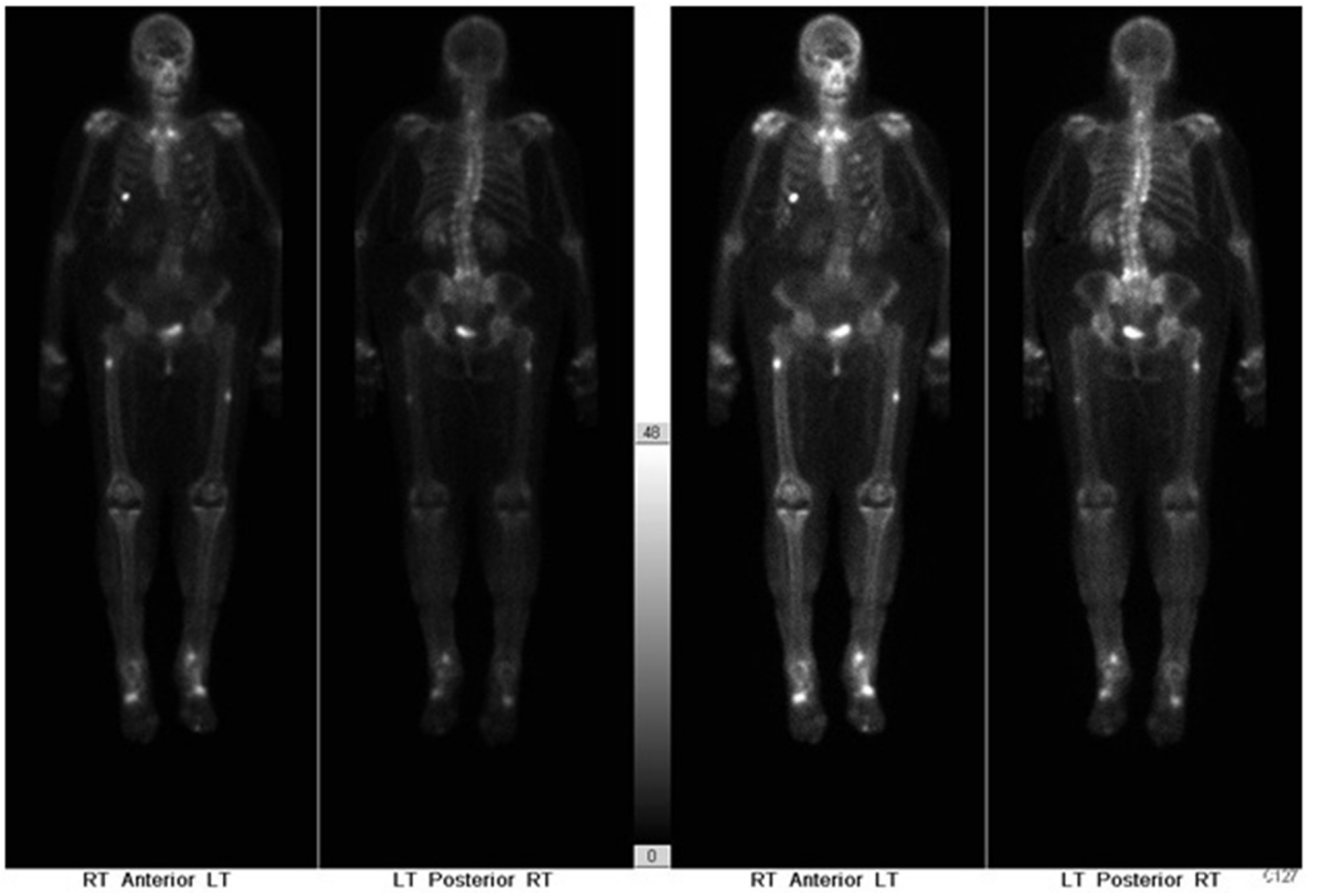


Figure 2. Whole-body bone scan showing focal activity on the lateral aspect of the proximal right femur and on the midshaft of the left femur.



Figure 3. Anteroposterior radiograph of right hip after fall causing AFF.

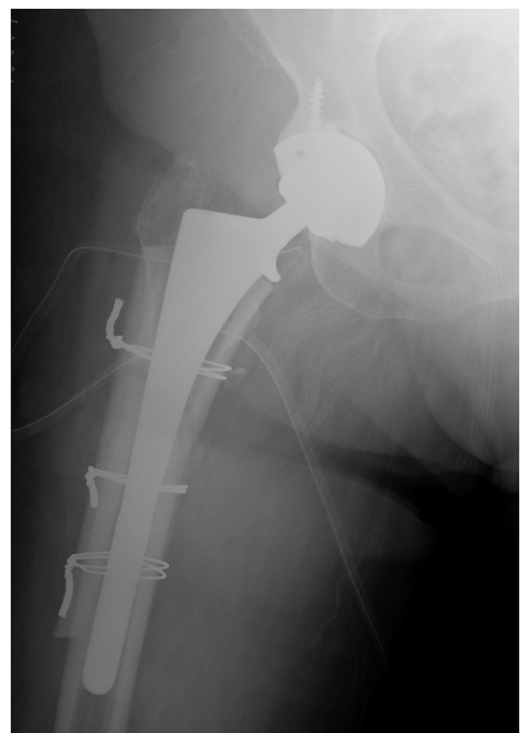


Figure 4. Postoperative anteroposterior radiograph of right hip.

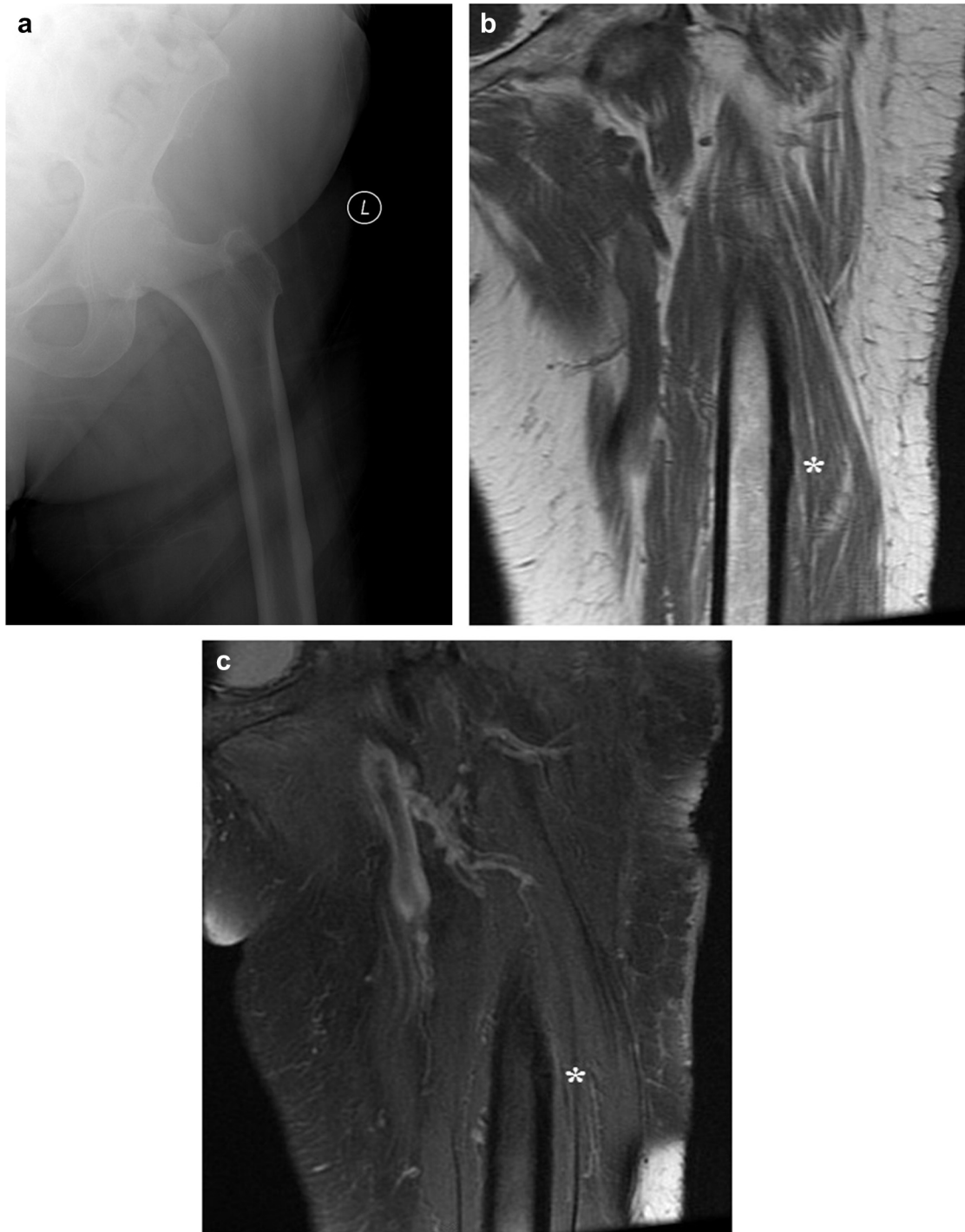


Figure 5. (a) Anteroposterior radiograph of left hip showing impending AFF. (b) T1 MRI of left hip showing impending AFF (at asterisk). (c) T2 MRI of left hip showing impending AFF (at asterisk).

Discussion

An AFF is defined as a transverse or short oblique non-comminuted fracture occurring between the subtrochanteric and supracondylar regions with no or only minor trauma [1]. The fracture is thought to be caused by modification of collagen cross-linking, accumulation of bone microdamage, increased mineralization or reduced heterogeneity of mineralization, change in bone metabolism, or reduced angiogenesis [12]. The mechanism by which BPs are thought to cause AFF is by inhibiting

osteoclast-mediated bone resorption, leading to inhibition of the normal bone remodeling process at the location of microfractures that normally occur [1,13]. Typical symptoms before the fracture are thigh or groin pain [4,14,15]. We classified both femoral fractures in the present case as AFFs because of the patient's history of long-term BP intake and the characteristic morphologic findings seen on the radiographs. In addition, the left impending femoral fracture was noted during the postoperative period while the patient was in a rehabilitation facility and without any instance of trauma.



Figure 6. Postoperative anteroposterior radiograph of left hip.

Both operative and nonoperative treatments of the impending AFF have been described. Operative treatment of the impending AFF uses intramedullary nailing or plating techniques, with excellent outcomes as shown by Egol et al. [16]. For nonoperative

treatment, the impending AFF is treated with partial weight-bearing plus or minus teriparatide treatment; results are mildly effective and many patients progress to a complete AFF [11]. Our case of complete and impending AFFs was treated with a long-stem uncemented THA with strut grafts on the right side and a long-stem cemented THA on the left side instead of internal fixation because the patient had concomitant symptomatic end-stage OA of both hips. We think that using the long-stem THA was a better choice to treat both pathologies simultaneously, when compared with completing internal fixation of the fractures and staging the total hips at a later time, because we were able to limit the number of procedures to 2 surgeries instead of 4 surgeries. To the best of our knowledge, there are no reports or guidelines concerning a specific course of action on the use of cemented or uncemented THA in face of atypical fractures. Our rationale for using an uncemented stem was the following: We used an uncemented stem with strut grafts on the right side to give extra support to the fracture site and enhance union without causing any leakage of cement into the fracture site which would have been the case if a cemented stem was used. The cemented stem was used on the left side to give extra support at the level of the impending AFF but, because it was not a complete fracture, the leakage of cement into the fracture site was not a concern.

Evidence-based studies, mostly large case series or retrospective studies, demonstrate an association between AFFs and BP use. However, large-scale epidemiologic studies have not confirmed this association [17]. It is important to note that these AFFs are rare and occur in a very small number of patients taking this type of treatment. We do not want to downplay the significant role of BPs in the treatment of osteoporosis [1]. Further research should include long-term, prospective observational studies examining the development of AFFs; specific data on bone biopsies and fracture healing in BP-treated patients; and further analyses of clinical, biomechanical, and genetic factors pertinent to the development of AFFs. Basic science investigations should examine the significance of cortical thickening, an unexplained feature of atypical fractures that may provide valuable insights into their pathomechanism.

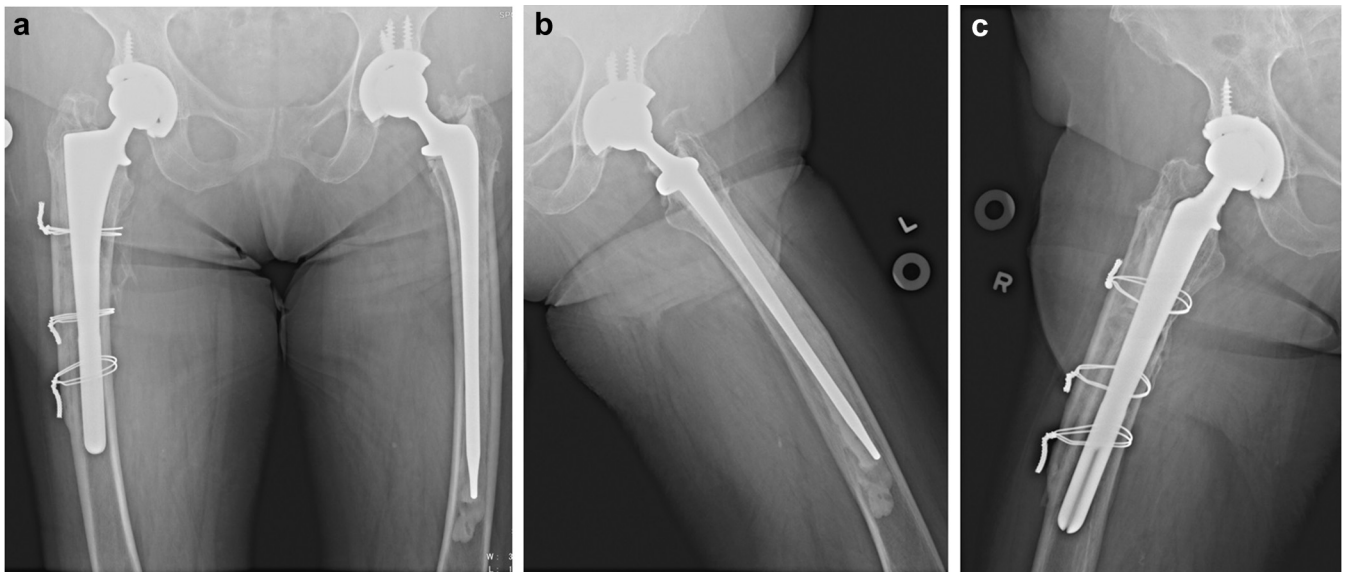


Figure 7. One-year follow-up radiographs: (a) anteroposterior pelvis, (b) lateral left hip, and (c) lateral right hip.

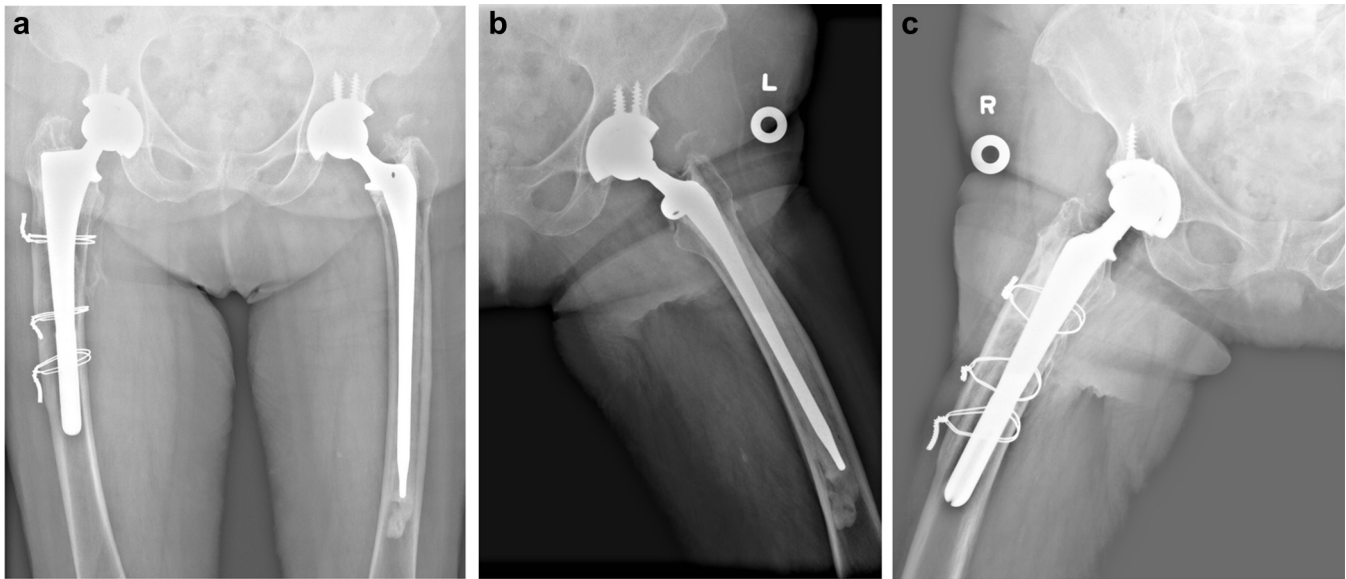


Figure 8. Two-year follow-up radiographs: (a) anteroposterior pelvis, (b) lateral left hip, and (c) lateral right hip.

Summary

We present the case of a patient who suffered a bilateral AFF in a short period of time around arthritic hips. This particular case illustrates that patients under prolonged BP therapy are at risk for AFFs even in the most controlled situations such as in the immediate postoperative period after a THA. This case also illustrates the short-term success of both cemented and uncemented total hip arthroplasties in the presence of these fractures.

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