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## Atypical femoral fracture in a patient with severely bowed femurs on short term bisphosphonates: A technical note

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### Abstract

**Background:** The incidence of atypical femoral fractures (AFF) is estimated at 70 to 100 per 100,000 patients. They are seen in patients who have undergone a prolonged course of bisphosphonate therapy. In one study, Asian women were found to have a more than 6-fold increase in the relative risk of AFF compared to Caucasians. It is also noticed that Asians have increased prevalence of bowing of the femur. This presents a challenge in the surgical management of AFF in Asians.

**Case Summary:** A 76-year-old woman presented to our institution after experiencing left lower limb weakness and fell subsequently. She sustained a left AFF. Radiographs and magnetic resonance imaging also revealed ongoing stress injury to her right femur. Both femurs were severely bowed. Prior to this, she has been undergoing alendronate therapy for 30 months for osteoporosis. She had no other fragility fractures. She claimed to have left thigh pain for 12 months before this presentation. She underwent plate osteosynthesis of her left femur spanning the femoral head and neck up to the supracondylar flare. She also had prophylactic fixation of her right femur with plate osteosynthesis.

**Conclusion:** A newer entity of AFF in patients on short term bisphosphonates use with severely bowed femurs should be recognised. Where there is difficulty in performing intramedullary nailing for these group of patients, whole length plating of the femur should be considered.

**Keywords:** Atypical fracture, bowed femurs, short term bisphosphonate, case report

### Introduction

The incidence of atypical femoral fractures (AFF) involving the diaphysis is estimated at 70 to 100 per 100,000 patients <sup>[1]</sup>. They are seen in patients who have undergone a prolonged course of bisphosphonate therapy <sup>[2, 3]</sup>. In one study, Asian women were found to have a more than 6-fold increase in the relative risk of AFF compared to Caucasians <sup>[4]</sup>. Interestingly, it is also noticed that Asians have increased bowing of the femur in both the coronal and sagittal planes <sup>[5]</sup>. This presents a challenge in the surgical management of AFF in Asians.

### Case report

#### Background

A 76-year-old woman who had suffered a left AFF presented to our facility in January 2020. She had a considerable medical history, including type II diabetes, hypertension, hyperlipidemia, and osteoporosis for which she received 30-month treatments with alendronate. She was pre-morbidly community ambulant, autonomous in her daily living activities, and aide-free.

A vague weakness in the patient's left lower limb caused her to stumble as she exited the kitchen, landing on her left side of the body and breaking her left femur. She had complained of left thigh soreness for at least a year prior to her fall. The pain was periodic, occurring mostly on "start-up", and occasionally at rest. She also admitted to having similar pain in her right thigh for a period of about 4 months.

Radiographs (Fig 1) revealed an atypical fracture of the left femoral shaft, with a short oblique fracture pattern, lateral cortical thickness, and no comminution. On the right femur, neither a fracture nor the "dreaded black line" were present, although there was a significant amount of periosteal response over the lateral cortex at the intersection of the middle and distal shafts (Fig 2a, b).



**Fig 1:** Radiographs showing atypical femoral fracture of the left femur shaft

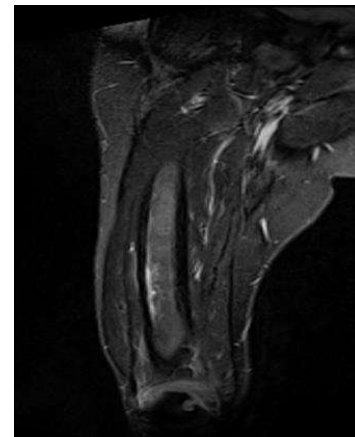


**Fig 2a:** Radiographs with a significant bend in the right femur shaft and endosteal and lateral cortical response

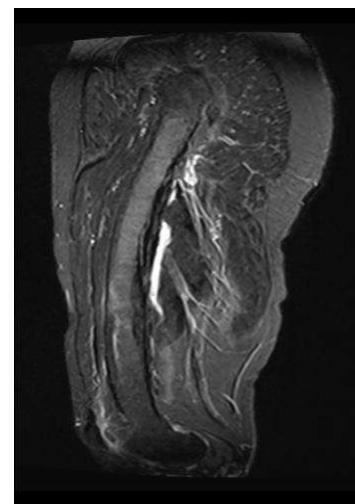


**Fig 2b:** Radiographs of the right femur shaft with a significant bow and endosteal and lateral cortical reaction

Anteriorly, both femurs showed an accentuated bow. The right femur (Fig 3a-b) underwent magnetic resonance imaging (MRI), which revealed periosteal reaction and oedema along the distal femur shaft, suggesting a continuing stress injury. According to the case definition published by the American Society for Bone and Mineral Research (ASBMR), the data indicated above met the requirements for the diagnosis of AFF.



**Fig 3a:** MRI scans of the right femoral shaft demonstrate bone oedema and periosteal-medullary inflammation, which are indicative of chronic stress damage



**Fig 3b:** MRI scans of the right femoral shaft indicate an ongoing stress injury because they exhibit bone oedema and periosteal-medullary inflammation

**Surgical Technique**

A surgical stabilization of the patient's left femur was recommended due to her favorable pre morbid condition. The right femur was used for preoperative templating. Her left femur's excessive bending made intramedullary nailing impractical, so she was scheduled for surgical fixation using a plate osteosynthesis.

The patient was lying supine on a radiolucent table during surgery while the patient was under general anesthesia. Two Schanz pins were positioned anteriorly on either side of the fracture to aid with reduction (and eventual compression at the fracture ends) after the fracture site was opened immediately laterally with elevation of the vastus lateralis. A lengthy plate (Zimmer NCB® Periprosthetic Femur System) was shaped and tunneled submuscularly onto the lateral surface of the femur after sufficient reduction. After that, screws were placed both proximally and distally. The variety of screw hole possibilities made implants the obvious choice. This was especially helpful because the femur's considerable bend prevents the plate from being completely shaped to match the bone. The implant also enables the insertion of screws into the femoral head and neck, which may provide additional defense against a proximal femoral fracture in the future (Fig 4a-b).



**Fig 4a:** Images from radiographs demonstrating a left atypical femoral fracture after extramedullary plate osteosynthesis and the restoration of the femoral bow



**Fig 4b:** Radiographs displaying a left atypical femoral fracture following extramedullary plate osteosynthesis with the restoration of the femoral bow

The patient had a smooth postoperative recovery in the ward, and 4 days later, she had preventive fixation of her right femur. The patient was positioned supine on a traction table during the procedure, which was carried out under general anesthesia.

The plate had a similar shape to her left side. The fixation was carried out using a minimally invasive submuscular method and a direct lateral approach. Once more, the same plate was employed, and screws ran proximally from the supracondylar region to the head-neck of the femur (Fig 5a-b).



**Fig 5a:** Radiographs showing prophylactic extramedullary plate osteosynthesis of right femur



**Fig 5b:** Radiographs showing prophylactic extramedullary plate osteosynthesis of right femur

The patient was permitted to fully bear weight on her right lower limb but not on her left lower limb without the use of

crutches for eight weeks. Six days after the second surgery, she was released to the community hospital for rehabilitation and made a good recovery.

At 15 weeks after her operation, she underwent a review in the outpatient clinics. On the left side of the body, radiographs revealed fracture union (Figure 6a-b).



**Fig 6a:** Radiographs showing left atypical femoral fracture 15 weeks post fixation



**Fig 6b:** Radiographs showing left atypical femoral fracture 15 weeks post fixation

She was ambulating independently then. She was last reviewed 2.5 years postoperatively. Full healing of the left femoral fracture was noted (Fig 7a-b)



**Fig 7a:** Radiographs showing full healing of left atypical femoral fracture 2.5 years post fixation



**Fig 7b:** Radiographs showing full healing of left atypical femoral fracture 2.5 years post fixation

**Discussion**

Currently, there are several studies discussing the associations between prolonged bisphosphonates usage and the increased risk of developing AFF [2, 3]. Generally, the median duration of bisphosphonate therapy in these patients stands at 7 years [6, 7]. Some studies reported that bisphosphonates use for more than 5 years were associated with an increased risk of diaphyseal femoral fractures [8, 9]. In contrast, our patient, had just been on bisphosphonates for 2.5 years. To develop an AFF is rather uncommon given

the short duration of therapy. To the best of our knowledge, there is paucity of literature exploring AFF in patients who underwent short term bisphosphonate therapy.

Rifai *et al.* [10] described a patient developing vague thigh symptoms 2 years after initiation of ibandronate. Their patient subsequently sustained an AFF on the right side with an incomplete fracture on the left approximately 5 years after therapy. They concluded that early determination of possible adverse events after starting bisphosphonates is imperative and raised the question of when further evaluations should be carried out in these patients. In our patient, thigh pain was experienced 1.5 years after initiation of bisphosphonates and a complete fracture was sustained a year later. This is alarming as it suggests the possibility that the adverse effects of bisphosphonates including suppression of bone turnover and accumulation of microfractures [10] may occur sooner than previously thought.

AFF have also been described in patients who never had bisphosphonate therapy [11]. The authors theorized possible causes such as metabolic abnormalities. However, our patient had denied any form of thigh symptoms prior to initiation of bisphosphonates. Also, baseline metabolic and biochemistry laboratory markers were largely normal, hence, we believe that bisphosphonates played a major role in causing her condition.

We hypothesize that with the increased anterolateral bowing of the femur, there is mechanical stress concentration in the anterolateral cortex of the femoral shaft. Also, with medial and posterior shift of the load axis by bowing, this results in increased distraction forces [12]. Together with the use of bisphosphonates causing accumulation of microdamage to bone, reduced heterogeneity of the organic matrix and mineral properties and increased advanced glycation end products with eventual deterioration of bone quality [7], the end result is the culmination of the atypical fracture.

The jury is still out with regards to the method of surgical fixation in AFF. Increasingly, the balance is swung towards favoring intramedullary fixation [13] with proponents claiming higher rates of postoperative implant fatigue failure with extramedullary plate osteosynthesis. Since bisphosphonates do not affect the initial inflammatory and callus formation phases but suppress the remodelling process of the bone by inhibiting the bone resorption of osteoclasts [14-16], delay in fracture healing from plate fixation is theorised [17].

Due to the significant femur bowing in our patient, preoperative templating with specialized Traumacad software ruled out the option of intramedullary nailing. Extramedullary plate osteosynthesis was chosen because of the risk of accidentally straightening the femur after nailing, which could result in an imbalance in limb length or femoral cortex perforation.

To provide protection for the relatively inert bone, we decided to span the proximal to distal femur, including the femoral head-neck, in our fixation. According to the ASBMR, atypical femoral fractures can happen anywhere from just proximal to the supracondylar flare to distal to the lesser trochanter. We decided to prevent a future recurrence of the fracture because doing so would make management more difficult. When using plate osteosynthesis, there is currently no agreement on whether to fixate these fractures in short or long segments.

As opposed to a distally based plate, we chose to protect the

femoral head-neck because we think that, should a fracture develop below the level of the plate, surgical repair of a distal based fracture will be simpler to execute. In circumstances where intramedullary nailing is challenging, Lee *et al.* have also described whole bone plating covering the entire length of the femur [18].

With the increased prevalence of femoral bowing in the Asian population [5], we are likely to see more of such cases similar to our patient. An index of suspicion should be maintained with patients on short term bisphosphonates therapy with concomitant bowed femurs. These patients should be followed up at closer intervals more regularly and alarm bells should ring should they complain of prodromal thigh pain.

### Conclusion

Increasingly, AFF due to prolonged bisphosphonates use are being encountered in clinical practice. A newer entity of AFF in patients on short term bisphosphonates use with severely bowed femurs should be recognized. Where it is difficult to perform intramedullary nailing for these group of patients, whole length plating of the femur should be considered.

### Conflict of Interest

Not available

### Financial Support

Not available

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