



Case Report

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Bilateral Simultaneous Atypical Femoral Fracture: A Case Report

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ABSTRACT

We report a rare case of bilateral subtrochanteric femoral fracture related to prolonged treatment with Alendronate. Bisphosphonates are the most common drugs used for treatment of osteoporosis and prevention of the risk of osteoporosis-related fractures. Atypical femoral fractures (AFF) have been reported in patients who have been on long term treatment with bisphosphonate especially Alendronate. our case was a 68- year- old woman who sustained bilateral subtroc- hanteric femoral fracture after a simple falling down. She had a past medical history of osteoporosis and had been on treatment with alendronate for 5 years. Radiographs showed bilateral fractures in femur subtrochanteric area. We decided to use double plating technique and we used angled blade plate and locking plate (LCP). Alendronate was discontinued and Teriparatide was started. Careful evaluation of hip or thigh pain in patients who are on prolonged courses of bisphosphonates is essential. Our method of fixation with double plating can be used in patients with poor bone quality.

Introduction

In the recent years, osteoporosis has been increased due to population changes and an increase in the elderly population. As a result, the risk of osteoporosis-related fractures has also increased which leads to much costs for the health care system [1].

Bisphosphonates are the most common

drugs that are widely used for the treatment of osteoporosis and many studies support their safety, cost effectiveness and their role in preventing osteoporosis-related fractures [2, 3]

Bisphosphonates have osteoclastic inhibition effect. They reduce bone resorption by inhibiting formation and aggregation of calcium phosphate crystals which increase bone density. They are useful in vertebral and nonvertebral osteoporotic fractures (such as hip

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fractures) by bone remodeling. [4-9]. Some adverse effects of bisphosphonates like esophageal diseases, atrial arrhythmia, renal toxicity, osteonecrosis of the jaw, myalgia [10] and concerns of bone delayed healing [11] have been reported.

Moreover, there have been reports of atypical femoral fractures (AFF) which occurred after long term use of Bisphosphonates [12]. Some studies reported that after long term use (i.e. more than 5 years) of these drugs, bone mineral density remains, but femoral neck density decreases [13].

Atypical femoral fractures associated with treatment with Bisphosphonates were first described in 2005. After that time, more reports indicated an increase in these types of fracture [14].

Atypical femoral fractures can occur after low energy traumas or without any traumas [15]. After 8 years of bisphosphonate use, the annual rate is 2 fractures per 100000 patients/years [16]. It is more related to alendronate use than other drugs of this family [17, 18].

Patients with atypical femoral fractures may have prolonged pain before diagnosis [19]. These fractures can occur in femur subtrochanteric area or in femur

diaphysis and can be complete or incomplete. They have some radiographic features; they are non-comminuted, transverse or short oblique fractures with medial spiking of the femoral cortex, local periosteal reaction exhibit, increased thickness of the lateral cortex, or they can have cortical beaking and diffuse cortical thickening [5, 20]. The relationship between this type of fracture and the use of bisphosphonates, and also the treatment options have always been an area of debate.

Case Presentation

The case of current study was a 68- years- old woman who was admitted to the hospital after a simple falling down from her own height. She had severe pain and was unable to move both of her legs, but neurovascular examination of lower limb was normal. She had a former medical history of osteoporosis. Therefore, she had been treated with alendronate 70 mg weekly for 5 years. Moreover, a systemic arterial hypertension was diagnosed 10 years ago and treated with losartan 25 mg daily. She had also hyperlipidemia which was diagnosed 7 years ago treated with atorvastatin 20 mg daily. In radiographic study, bilateral subtrochanteric femoral fracture was shown (Fig. 1). Both fractures were the same transverse lines with no comminution. A little medial spike was in left femoral fracture. She



Fig. 1. Bilaterally Subtrochanteric Atypical Fracture

underwent surgery for open reduction and internal fixation (ORIF) with 95 degree angled blade plate and augmentation with anterior 3.5 mm locking plate (LCP), due to concern about long time needed for

union in subtrochanteric area especially in atypical form (Figs. 3,4). Patient was discharged from hospital after 48 hours with anticoagulant treatment and no weight bearing.

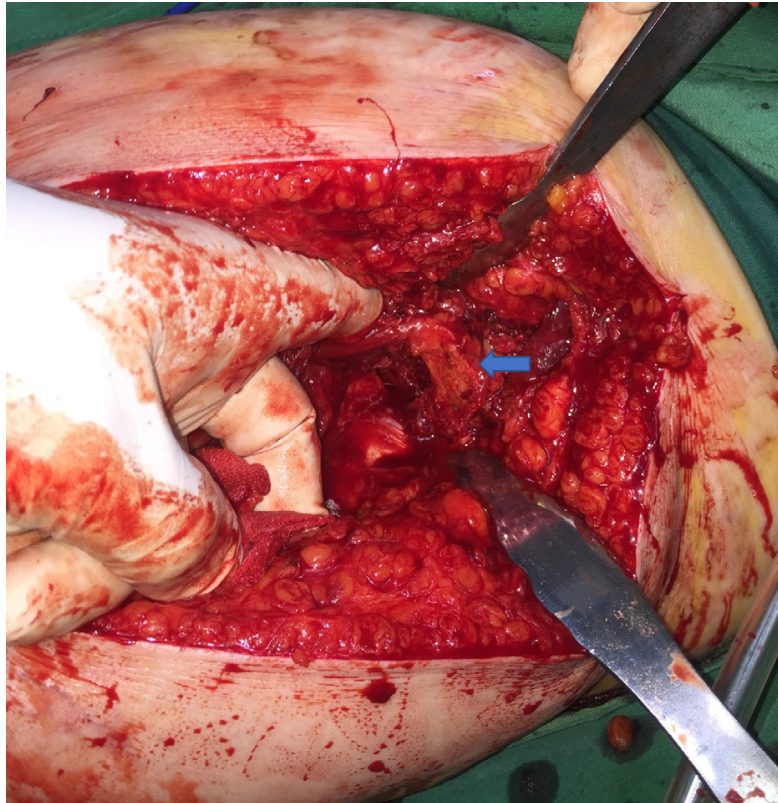


Fig. 2. Lateral cortex thickening due to bisphosphonate use



Fig. 3. Anteroposterior postoperative radiograph

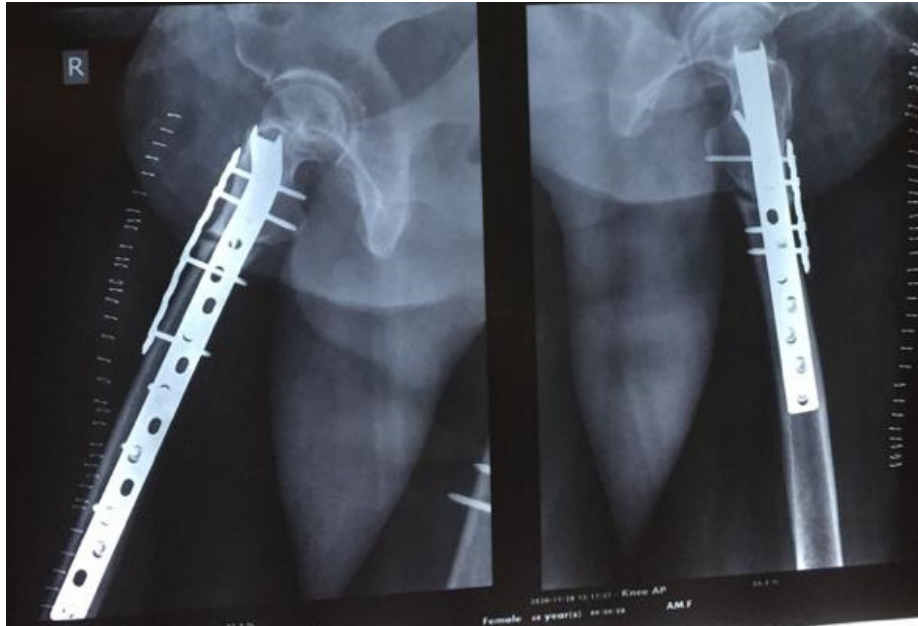


Fig. 4. Lateral postoperative radiograph

Discussion

We report a rare case of simultaneous bilateral subtrochanteric femoral fracture due to long term alendronate use.

As we know the most common risk factor for fracture of femur and spine in elderly population is osteoporosis and for preventing this, bisphosphonates are widely used by this group of patients especially in postmenopausal women [21, 22]. Bisphosphonates have an analogue effect on inorganic pyrophosphates and they inhibit osteoclast activity and bone resorption [23]. These drugs have two subtypes: 1- non nitrogenated such as clodronate, etidronate and tiludronate, and 2- nitrogenated like alendronate, ibandronate, risedronate, pamidronate and zoledronate [24]. One of the concerns of the wide use of bisphosphonates is atypical femoral fracture [25]. Although the exact mechanism of AFF is still unclear, there are theories that inhibiting osteoclast mediated bone resorption causes slow bone turnover which can result in microdamages that lead to AFF [26, 27]. As a result, there is a relation between geometry of femur and AFF and its location [28]. In some studies, possible risk factors for AFF occurring in bisphosphonate users were osteomalacia, vitamin D deficiency, curved femurs and slow bone turnover [4]. In other studies, genetic trait, Asian ethnicity, altered collagen cross linking and reduced vascularity have been associated with AFF [29, 30]. In a study by Edwards et al., AFF was associated with rheumatoid arthritis and hypophosphatasia [4]

and in the other study by Gedmintas et al., medications other than bisphosphonates were considered related to AFF. Some of these medications were corticosteroids, hormone replacement therapy, etanercept and proton pump inhibitors [20]. In addition, statins were suggested as a cause of AFF but there was no statistical significance [31]. Radiographic features of AFF are fracture line originating at lateral cortex, transverse fracture line, beaking medial spike, and non-comminuted fractures [32]. Beaking is a specific finding for AFF [33]. In a study done by Rosenberg et al., lateral cortical thickening, transverse fracture and medial spike were predictors for bisphosphonates fractures [34]. In some reports, fractures happened bilaterally [35].

Incidence of AFF is 7.8/100000 person-years in patients over 60 years old. In the studies, by every two years of bisphosphonates use, the incidence increases 2/100000 person-years and for every 8 years of exposure, this number is 78/100000 person-years [36].

Cessation of bisphosphonates can decrease the risk of AFF rapidly, and after diagnosis of AFF, they should be discontinued [37, 38]. In incomplete fractures, cessation of bisphosphonates and conservative treatment can be chosen, patient should limit the load on affected limb and use crutches. So, if there is no evidence of fracture healing after 3 months, the patient should undergo surgery [36]. Teriparatide is a recombinant form of parathyroid hormone and it has healing effects on bone, so it can be used in this type of fracture [39].

Treatment of choice in AFF is open reduction and internal fixation [36]. These fractures can be treated by intramedullary rods and plates and screws [40]. In many studies, intramedullary fixation is a selective treatment. Moreover, there are studies that advocate plates and screws are ineffective [41]. But in Black et al.'s article, they prefer extramedullary fixation instead of intramedullary, because these fractures usually happen in patients who use bisphosphonates and these drugs can reduce bone remodeling by osteoclast inhibition that results in poor intramembranous fracture healing [19].

Our patient was a case of osteoporosis on long term bisphosphonate treatment. she came to the hospital with simultaneous bilateral atypical subtrochanteric femoral fracture and poor bone quality, so we decided to use double plating technique and we used angled blade plate and LCP. Alendronate was Discontinued and Teriparatide (Cinopar®, CinnaGen Co.) was started.

Ethical Considerations

Compliance with ethical guidelines

All ethical principles are considered in this article. The participants were informed of the purpose of the research and its implementation stages. They were also assured about the confidentiality of their information.

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Authors' contributions

All authors equally contributed in preparing this article.

Conflict of interest

Authors declare that there is no conflict of interest.

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