The 5-year follow up of a cortical stress fracture resulting in a spontaneous atypical subtrochanteric femoral fracture in a female patient with severe osteoporosis and bisphosphonate therapy over 15 years

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Introduction

suppression of bone turnover by the antiresorptive effect of BP has been a therapeutic standard in the management of osteoporosis for the last few decades. Recently, the possible relationship between long term treatment with BP and the incidence of atypical femur shaft fractures has been controversially discussed in numerous case reports and population based studies [2–5]. In the most recently published Task Force Report of ASBMR [1] all reports on this topic had been reviewed. There is evidence that prolonged reduction of bone remodelling is leading to microcracks and suppression of repair mechanisms. These
microcracks occur predominantly in cortical bone in areas with high tension like the femoral shaft. However, an absolute risk of atypical fractures in association with BP for an individual person has not been proven so far.

Case report

A 64-year-old woman with osteoporosis was admitted to the St. Vincent Hospital Vienna in January 2000 because of persistent and increasing pain in the right femur for 3 months. Her anamnesis showed no evidence of any trauma. The pain caused severe disability in walking and standing. The symptoms did not respond adequately to analgetic therapy with NSAIDs and oral morphins.

After an early menopause at the age of 45 the patient was treated with hormone replacement therapy (HRT). At the time of her first admission to our hospital she had already received oral BP over 10 years (cycling treatment with Etidronate 1990–1995, Alendronate 70 mg weekly 1995–2000).

The medical history did not reveal any other disease; except for an early menopause no other clinical risk factor for osteoporosis according to the FRAX algorithm could be explored.

The X-ray (Fig. 1) showed a horizontal compact formation of bone tissue in the subtrochanteric region corresponding to a microfracture in the cortical area. Any other cause of the bone lesion, e.g. inflammatory changes, could be excluded by MRI. We performed a fine needle biopsy which showed osteoporotic cancellous bone and post-inflammatory morphology with myelofibrosis and lymphatic cells, malign changes could be excluded.

Repeated laboratory findings did not show any evidence of systemic or metabolic diseases. Serum osteocalcin (4.6 ng/ml, normal range 18.4–41.4 ng/ml) was suppressed as a consequence of the long-term BP treatment. There was a transient slight increase of PTH (80.8 pg/ml, normal range 12.0–72.0 pg/ml) most likely due to a mild 25-OH vitamin D deficiency (29.4 ng/ml), which was corrected by CA/D supplementation after 6 weeks.

With respect to the significantly decreased BMD the peroral BP medication was continued since the alternative treatment with Calcitonin had to be stopped after 5 days because of an anaphylactic reaction with palmo-plantar-erythema. Additional antiphlogistic and analgetic treatment was initiated with 75 mg Diclofenac and 500 mg Mefenaminacid.

After another period of 5 years under continuous treatment with oral BP (Alendronate 70 mg weekly, 2000–2005) and stable conditions regarding her BMD values of the spine and hip (Table 1) the patient was admitted again with increasing pain in the right femur, resistant to analgetics. The X-ray and computed tomography showed a progression of the microcrack in the subtrochanteric region with disruption of the corticalis contour similar to a manifest stress fracture (Fig. 2), in addition a new vertebral fracture of L4 was found without any preceding trauma. In the bone scan (Fig. 3) an accumulation of Technetium (Tc99) in the corresponding region of the right femur was detected. (Fig. 4)

To prevent a complete subtrochanteric fracture, the patient was transferred to an emergency hospital in Vienna for surgical intervention. After refusing any intervention,
4 weeks later a spontaneous complete atraumatic fracture occurred and a gamma nail had to be implanted (Fig. 5).

Discussion

Stress fractures in the cortical part of bone are observed in areas with typical high tensional stress in the proximal femoral shaft. Those changes in the femoral shaft are thought to be possible prodromal changes followed consequently by complete fractures.

The recently published Task Force Report of ASMBR [1] identified major and minor features to assist in finding and reporting these atypical fractures. According to these, it seems that our patient suffered from an atypical femoral fracture, which was located in the subtrochanteric region and occurred spontaneously, not associated with any trauma. The fracture had a short oblique configuration, was noncomminuted and crossed both cortices. On admission to our clinic the patient had been under BP medication for 10 years. Imaging techniques of the right femur showed typical early morphologic changes of bone structure in the cortical area of the subtrochanteric region. Nevertheless, our patient had an incident vertebral fracture of L4 under BP treatment while having stable BMD values.

It is well accepted and has been proven that the incidence of hip-fractures has declined since BP were approved for use. There are incongruent data of the incidence of subtrochanteric fractures or shaft fractures over the same period. In a recent publication Schilcher et al. [2] showed in a population-based nationwide analysis in Sweden a small absolute risk for atypical fractures but a high prevalence of BP use among those patients. In this study, the risk to sustain an atypical fracture increased with the duration of BP treatment and decreased after withdrawal, which could favour intermittent therapy. It was assumed that changes in the cortical region are common in patients with atypical fractures, especially at the lateral site. In our patient a microcrack and stress fracture at the lateral corticalis had been observed 5 years prior to the occurrence of a spontaneous complete fracture of the femoral shaft.

In general, atypical femoral fractures are rare in BP-treated patients but there seems to be an increase with persistent exposure to BP. It is important to note that atypical fractures have been reported also in patients without any exposure to BP. According to epidemiologic data, subtrochanteric and femoral shaft fractures follow an age and sex distribution similar to osteoporotic fractures.

The risk-benefit ratio clearly favors BP treatment in women with high risk of fracture [7], however, more data are needed to verify the evidence of increased risk under prolonged therapy.

The conclusions based on the chronology of the morphologic changes being responsible for the development of the atypical femoral fracture are as follows.

After 10 years of BP treatment our patient showed a micro crack in the cortical subtrochanteric region of the right femoral shaft, which has to be interpreted retrospectively as an early sign of bone structure insufficiency. After another 5 years of BP medication the further progression of
the lesion resulted in an atraumatic complete fracture. With the current state-of-the-art knowledge we would have made a different decision because of more therapeutic options in the management of osteoporosis and the evidence of enhanced fracture healing caused by osteoinductive agents. Irrespective of the surgical intervention of the manifest stress fracture to prevent a complete fracture, the patient would have benefitted most likely from an osteoanabolic treatment with PTH analogs [8].

This is so far the first Austrian report of an atypical subtrochanteric femoral fracture which is possibly related to long term treatment with BP. Therefore, an initiative to establish a population-based registry evaluating hip, subtrochanteric and diaphyseal fractures in Austria is being started to compare hip fracture incidence in BP users to that in non-users.

Conflict of interest
The authors declare that there is no conflict of interest.

References