We present the case of a 34-year-old woman with a bilateral identical stress fracture of the distal fibula with a two-year interval. Stress fractures in young patients are most likely fatigue fractures caused by high physical activity, in this report however this was not the case but underlying osteopenia was detected during further exploration.

Keywords: Fatigue fracture; Postpartum; Fibula; Osteoporosis

Case Report

A 34-year-old woman with a clean medical history presented in 2009 with pain in the left lateral malleolus and foot. Four weeks earlier she gave birth to her second child after an uncomplicated pregnancy without excessive weight gain nor physical activities. The pain started two weeks before delivery. There was no history of trauma. Application of ice and use of anti-inflammatory drugs did not relieve the pain. Upon clinical examination tenderness and swelling were found on the left lateral malleolus but no pain could be provoked posterior or inferior to this region. Full weight bearing on the left foot was not possible, resulting in abnormal gait.

Radiographs of the ankle showed a distal transverse fracture of the fibula with postero-lateral displacement (Figure 1).

Based on the displacement, the presence of persisting pain and the limited time available for recovery due to the new-born infant, surgical osteosynthesis was preferred over conservative treatment. During surgery additional lyophilized bone grafting was necessary after debridement of the fracture. Three cortical screws were used proximally and two locking screws distally to fixate the plate (Figure 2). During the revalidation period she used a walker boot for four weeks. Two months postoperatively some mild pain persisted over the lateral malleolus but the patient could resume her daily activities including her work as social helper. This pain resolved spontaneously two weeks later.

Only two years later (2011) the patient consulted for the same complaint but now at the right ankle. Remarkably she just had her third child two months ago at 32 weeks of gestational age and was still breast-feeding. Before the delivery she rested in bed for six weeks. Mild swelling and tenderness were found at the joint between fibula and talus. Radiographs showed a transverse fracture of the distal fibula with mild postero-lateral displacement (Figure 3). Because the previous osteosynthesis was successful, the same treatment was applied now. The fracture healed well despite a superficial wound infection and the patient could go back her normal level of activity.
Bone mineral density, obtained using dual energy x-ray absorptiometry (DEXA, QDR-1000, Hologic inc.), showed osteopenia at L1-L4 (T=-1.46), at the femoral neck (T=-2.39), at the total hip joint (T=-2.06) and osteoporosis at Ward’s triangle (T=-3.04) two weeks after the second osteosynthesis. Blood tests were performed at the same time and showed no abnormal levels of ions, parathyroid hormone (PTH) and vitamin D. She was a non-smoker and seldom used alcohol. After surgery she was referred to the osteoporosis clinic where daily calcium and vitamin D supplementation were prescribed accompanied by weekly oral risdonate.

Discussion

Stress fractures are first described by Breithaupt, a doctor of the Prussian army in 1855. Since then, they are commonly seen in the military but in the past decennia they are more and more diagnosed in athletes and osteoporotic patients. This pathology can be divided into two types based on the underlying mechanism: fatigue and insufficiency fractures. Fatigue fractures are seen in athletes and other physically active people when abnormal repetitive stress is applied to normal bone. Insufficiency fractures arise when abnormal bone is subjected to a normal amount of stress.

The fracture’s location is related to the sport performed: metatarsal “march” fracture, proximal fibula “jumpers” fracture and distal fibula “runners” fracture [1]. The fibula carries between 6.4% and 16.7% of the load applied to the leg, the remaining load is carried by the tibia, making this the most common site of stress fractures (49.1%) [2,3]. In athletes only 6.6% of all stress fractures occur in the fibula and in this bone the distal third is the most commonly affected site [3-5]. Wang et al argued that this is due to the interosseous membrane. This membrane directs the axial load onto to the distal third of the fibula. Fractures at this level have been classified into two types by Burrows in 1948 [7]. In young male athletes the fracture site is located five to six cm proximal to the tip of the lateral malleolus while in middle-aged females this is approximately three to four centimetres from the tip. The can be explained by the difference in bone composition. The bone of the higher distal part is mainly cortical while the lower distal part mainly consists of cancellous bone which is more susceptible to the increased bone turnover in osteoporosis. The first type is more common and is also called a “runners” fracture [8]. The latter type is seen in osteoporotic women and is mostly an insufficiency fracture.

Bilateral fibular fatigue fractures were described in eight cases [9]. To our knowledge, Kazimoglu et al. was the only one to report on a case of bilateral fibular insufficiency fractures [10]. He described the case of a 54-year old woman diagnosed with, and treated for osteoporosis who developed bilateral ankle pain after a period of increased walking. Bilateral distal insufficiency fractures were found on standard radiographs, CT and technetium-99 m bones scan. The locations of the fractures were identical to ones found in our patient but no dislocation was present in this case. The patient was successfully treated with rest, a four week cast immobilisation and restricted weight bearing for six weeks.

Insufficiency fractures, which imply poor bone quality, are often caused by osteoporosis, Paget’s disease and rickets [8]. In our patient the diagnosis of osteoporosis could not be confirmed by DEXA (hip and vertebral values were greater than -2.5) but we could withhold severe osteopenia at the level of the hip. A z-score of -3.04 was measured in Ward’s triangle, but only hip and vertebral values can be used in the diagnosis of osteoporosis [11]. Based on these data and the location of the fractures (3cm above the tip of the lateral malleolus), both fractures were classified as insufficiency type stress fractures.

Standard treatment for this type of stress fractures consists of relative rest for three to eight weeks. Cast immobilisation and pneumatic ankle braces have also shown to be effective. Crutches and partial weight bearing are only used in selected cases.

However when a fracture line is visible or signs of delayed union are present or when there is some fracture displacement, surgical intervention with plate and screw fixation is necessary [8]. In this case the two fractures were displaced postero-laterally so internal fixation was performed using a plate, three cortical and two locking screws.

The reason for the osteopenia found in our patient could be explained by the relationship between pregnancies and the development of stress fractures.

During pregnancy the foetal calcium demands are partially compensated by increased resorption in the bowel. Eighty per cent of all maternal-foetal calcium transfer occurs during the third trimester [8]. Studies have shown a decrease of 4-5% of lumbar spine bone mineral density during pregnancy [13]. In the lactation period skeletal stores are used to provide calcium to the child via breast milk. Renal preservation also contributes to the blood calcium stores. Daily, 280 to 400mg of calcium are lost through breast milk. This results in a decrease of BMD of 1-3% per month. In comparison to postmenopausal osteoporosis in which an average of 1-3% per year is lost, this rate is extremely high. Studies have shown that calcium supplementation during lactation does not prevent this [14]. The mechanism behind this bone loss is partly explained by a decreased oestrogen level but other key molecules like parathyroid hormone related protein (PTHrP) possibly play a role. In postmenopausal osteoporosis there is a loss of calcium via the kidneys but during lactation PTHrP reduces renal calcium loss. Parathyroid hormone (PTH) itself is low in both conditions [12].

Another factor that may contribute to the osteopenia apart from breast-feeding is disuse osteoporosis because of the 6 weeks bed rest at the end of the pregnancy. The bone is subjected to three kinds of mechanical stress: gravity-related weight bearing, ground reaction forces and dynamic load generated by muscle contractions. The last two of these are reduced when patients are subjected to a prolonged resting period. Areas rich of cancellous bone are very sensitive to decreased activity. A decrease of bone mass of 2% at the distal tibia and 1% at the distal femur have been described after only 35 days of...
bed rest. Cortical bone decreases by less than 2% after 90 days [15]. The loss of bone mass after 6 weeks bed rest can be significant enough to have an impact on the BMD of this patient.

In the case of lactation osteoporosis or osteopenia, weaning from breast-feeding, calcium and vitamin D improve BMD but usually there is no return to normal levels. An average increase of 6% at 8-18 months and 9.6% at 2-4 years of spinal BMD is to be expected with this therapy [16]. Adding antiresorptive agents (bisphosphonates) results in a 23% increase in two years’ time [13]. A treatment period of five years followed by bone densitometry seems a reasonable approach for this condition.

**Conclusion**

This rare case of bilateral insufficiency fractures of the distal fibula is most likely caused by osteopenia during the pregnancy and lactation period combined with lowered BMD due to disuse in the period before the second fracture. Surgical treatment with plate and screw fixation was necessary because mild displacement of the fracture was present, resulting in an excellent outcome. To correct the underlying osteopenia, treatment with calcium, vitamin D and risedronate were started for at least five years.

**References**