


Bilateral femoral neck fractures resulting from pregnancy-associated osteoporosis showed bone marrow edema on magnetic resonance imaging

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Abstract

Femoral neck fractures resulting from pregnancy-associated osteoporosis is a rare condition. Herein, we report an undoubted case of pregnancy-associated osteoporosis in a 38-year-old primiparous patient with pre-existing anorexia nervosa who suffered bilateral femoral neck fractures in the third trimester and early post-partum period. Magnetic resonance imaging revealed femoral neck fractures as well as diffuse marrow edema involving both femoral heads, which are considered under ordinary circumstances as characteristic imaging findings of transient osteoporosis of the hip. Based on our experience, we propose that pregnancy-associated osteoporosis might be present in femoral neck fractures attributed to transient osteoporosis of the hip in pregnancy. Conversely, bone status should be carefully and accurately estimated in cases of potential transient osteoporosis of the hip in pregnancy to reduce future fracture risk.

Key words: anorexia nervosa, bone marrow edema, femoral neck fracture, osteoporosis, pregnancy.

Introduction

Musculoskeletal complaints are very common in pregnancy, but cases of non-traumatic bone fracture are extremely rare. Curiously, while cases of vertebral fracture are generally diagnosed as pregnancy-associated osteoporosis (PAO), cases of femoral neck fracture have frequently been reported as transient osteoporosis of the hip (TOH) in pregnancy, and general bone status in these cases has often been poorly assessed.^{1–3}

Pregnancy-associated osteoporosis is a fragility fracture that rarely occurs non-traumatically during pregnancy and lactation, during which time the maternal skeleton could be threatened.⁴ Femoral neck fracture resulting from PAO is a rare complication during pregnancy, as vertebral fractures are the most common effect.^{1,4,5} About 80% of calcium provided to the fetus

by term is accreted during the third trimester when the fetal skeleton undergoes rapid mineralization. Generally, this demand for calcium is largely met by increased efficacy of intestinal calcium absorption. However, if the maternal intake of calcium is insufficient, the maternal skeleton will undergo resorption during the third trimester, and such bone loss in combination with the increased body weight during this period may threaten the weight-bearing joints. Women with already low bone mineral density (BMD) because of reduced bone mass conditions prior to pregnancy would thus be at higher risk of fracture in late pregnancy. Because most affected women appear otherwise healthy, preceding BMD and biochemical data are seldom available, and this lack of data is likely to obscure the pathogenesis of PAO.⁴

Another disease that rarely causes non-traumatic fracture is TOH. TOH affects men more frequently than

Received: October 7 2016.

Accepted: January 22 2017.

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women, but pregnancy-associated cases are common.^{6–8} Transient osteoporosis is characterized by severe joint pain arising in the absence of related trauma. This pathology most commonly involves the hip, followed by other joints of the lower extremities.⁹ Plain radiographs show radiolucency of the affected joints, reflecting regional osteopenia,^{2,8,9} but magnetic resonance imaging (MRI) is currently the diagnostic modality of choice.^{2,6,7,10,11} Bone marrow edema (BME) on MRI is characteristic and is detected with high sensitivity.

Herein, we report a case of PAO in a patient with pre-existing anorexia nervosa (AN) who suffered bilateral femoral neck fractures in the third trimester and early post-partum period. This appears to be the first report to show characteristic imaging findings of TOH in a patient with PAO.

Case report

A 38-year-old, gravida 0, para 0 woman presented with severe pain in both hip joints. In terms of past history, she had developed an eating disorder at 18 years old and amenorrhea at 20 years old. She consulted a gynecologist and psychiatrist, both of whom diagnosed AN, but she only continued follow-up with these doctors for two years. The patient was 157 cm tall, and her weight and body mass index (BMI) were 33 kg and 13.4 kg/m², respectively. Several years after the onset of AN, her appetite spontaneously recovered and she gradually regained weight. Menstruation resumed at 31 years old, at a weight of 40 kg, and her menstrual cycles had thereafter remained regular. She married at 34

and became pregnant spontaneously at 37 (weight 40 kg, BMI 16.2 kg/m²).

The patient had no preceding history of trauma, fever, disease other than AN, smoking or alcohol abuse. Family history revealed that her mother had developed vertebral compression fractures resulting from osteoporosis at 60 years of age.

Pain localized over the left hip joint began at 28 weeks of gestation, of unknown cause. This pain progressively increased to involve both hip joints.

In consultation with an orthopedician at 32 weeks of gestation after hip pain continued to increase, left hip range of motion was 0–90° for flexion and 20° for external rotation, limited by pain, whereas rotation of the right hip showed no limitation. The Patrick test showed positive results in the left leg and negative results in the right. No imaging was performed because of the potential risk to the unborn child. Osteoarthritis of the hip and probable acetabular dysplasia were suspected, and conservative treatment was followed.

At 35 weeks and five days of gestation, she was admitted to our hospital with difficulty walking. At this point, she weighed 48.2 kg. Serum 1,25(OH)2D3 and parathyroid hormone levels were not measured, but other laboratory investigations revealed no abnormalities. MRI of the hips showed a fracture of the left femoral neck as well as diffuse BME involving both femoral heads. T1-weighted imaging revealed decreased intensity of the bone marrow (Fig. 1a), while short-tau inversion recovery T2-weighted imaging revealed a matching increase in intensity (Fig. 1b). Epiphyseal stress fracture, which is associated with avascular necrosis of the femoral head, was not observed. Cesarean section was performed two days after admission and a healthy 2720 g girl was

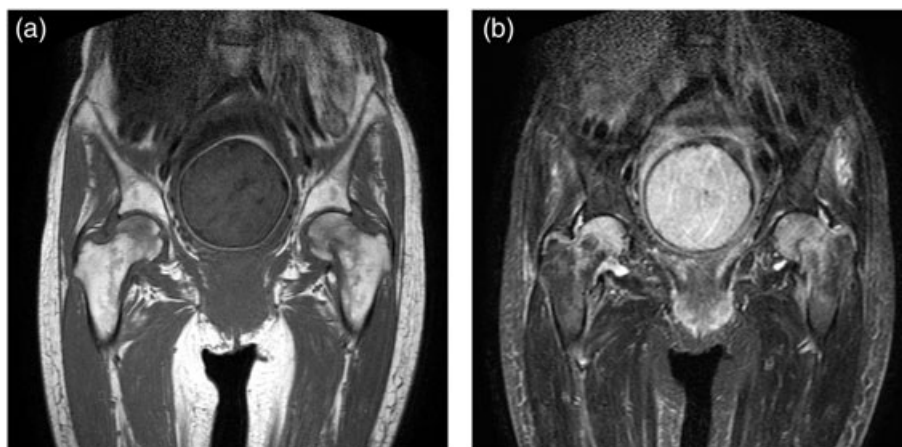


Figure 1 Signal intensity is (a) decreased in bilateral proximal femora on T1-weighted imaging and (b) increased on short-tau inversion recovery T2-weighted imaging.

delivered. Surgical intervention for the fracture was scheduled for a week after cesarean section, but the right hip pain worsened on the third day. On post-partum day 5, dual-energy X-ray absorptiometry of lumbar vertebrae 2–4 showed a T score of -3.5 , a Z score of -3.5 and a BMD of 0.696 g/cm^2 , representing osteoporosis according to World Health Organization guidelines. Radiography of the pelvis performed the same day showed bilateral femoral neck fractures (Fig. 2).

The patient immediately underwent open reduction and cannulated screw fixation of both fractures, followed by administration of teriparatide at $20 \mu\text{g/day}$ and calcium lactate at 2 g/day , considering that she was still of reproductive age. The neonate was bottle-fed. At five months post-operatively, the patient was able to walk without pain.

Discussion

This case appears unique in demonstrating BME involving both femoral heads on MRI in a patient who suffered from femoral neck fractures resulting from PAO. BME was already present before the fracture on the right, indicating that the BME had not resulted from fractures. Based on our experience, we propose that PAO might potentially be present in cases of femoral neck fracture reported as TOH in pregnancy.

Our patient could have been diagnosed as suffering from PAO for a variety of reasons. She showed various risk factors for PAO, including previous AN, long-term low body weight and a positive family history.⁴



Figure 2 Radiography of the hips shows fractures of the bilateral femoral necks.

Osteoporosis is a significant consequence of AN. A negative correlation has been found between spinal bone density and duration of amenorrhea as an indicator of the severity of estrogen deficiency in AN women.¹² A longitudinal study identified the critical BMI for a positive increase in BMD as $16.4 \pm 0.3 \text{ kg/m}^2$, suggesting that BMD does not increase under conditions of very low body weight.¹³ In addition, the reduced BMD in AN women is stable over time, with no rapid reversal after weight recovery, even after resumption of menses.¹⁴ Because body weight and BMI in our patient at the time she became pregnant were 40 kg and 16.2 kg/m^2 , respectively, we speculate the presence of general low BMD that remained unidentified before pregnancy. In addition, the extremely low spinal BMD appearing in the early post-partum period suggested preceding generalized osteoporosis.

Our opinion that TOH in pregnancy might occur in association with preceding general low BMD is also supported by epidemiological data, in that bilateral disease is rare in TOH overall, but not in pregnancy cases.^{6,8} Although the pathogenesis of TOH remains poorly defined, and has classically been related to regional periarticular disorders,^{4,9} recent investigations have indicated that general low BMD is potentially associated with the occurrence of TOH. Bone densitometry at the lumbar spine, an asymptomatic site, has quantitatively demonstrated generalized osteoporosis in TOH in pregnancy.^{3,10} MRI studies have indicated that TOH in both sexes is highly associated with generalized osteoporosis, as well as regional osteopenia.^{6,11}

Our report highlights a clinically rare complication that clinicians should be aware of. Because musculoskeletal complaints are common and radiography is often avoided during pregnancy, PAO might be underreported. Severe hip pain in lean pregnant women during the third trimester should prompt clinicians to perform an MRI. An MRI is considered most useful as a non-invasive modality, while the benefits of radiography might outweigh the minimal risk to the fetus in this period.

Potential factors contributing to bone density loss in cases of femoral neck fracture during pregnancy that show characteristic findings of TOH should be suspected, whereas most cases of TOH resolve spontaneously without sequelae under conservative management. In potential cases of TOH in pregnancy, bone status should be estimated carefully and accurately to reduce future fracture risk.

Women with AN who fail to gain sufficient weight before pregnancy are at high risk of fracture during

pregnancy and lactation. In particular, osteopenic status in young patients who fail to attain an optimal peak bone mass should be better treated before pregnancy. In addition to AN patients, a large number of women are potentially vulnerable because of the common desire to lose weight in the young populations of developed countries, even when BMI values are normal or low. Low BMI in the healthy population is associated with increased risk of femoral neck fracture, even after adjusting for BMD.¹⁵

Disclosure

None declared.

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