CASE REPORT

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A Postpartum Woman Diagnosed with Multiple Vertebral Fragility Fractures Due to Pregnancy and Lactation-Associated Osteoporosis

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ABSTRACT Pregnancy and lactation-associated osteoporosis (PLO) is a rare condition diagnosed with vertebral fractures detected in late pregnancy and early postpartum periods. The incidence of PLO is reported to be 0.4 in 100 000 women. Maternal bone resorption and absorption of calcium from the intestines are increased in order to ensure adequate calcium transfer to the fetus, 80% of which is in the last trimester. Bone mineral density decreases during lactation and is associated with high bone turnover. Although various factors have been suggested to be responsible, the cause and etiopathogenesis of osteoporosis associated with pregnancy and lactation have not been fully elucidated. Herein, we reported a 28 year-old primigravid woman without risk factors for secondary osteoporosis diagnosed with vertebral fractures at postpartum third month.

Keywords: Pregnancy; osteoporosis; lactation

Pregnancy and lactation-related osteoporosis is a rare form of osteoporosis. Diagnosis is made mostly with the detection of vertebral fractures in patients who present with low back and back pain in the last trimester and postpartum first months. During pregnancy various changes occur in the mother's bone and mineral metabolism to provide calcium transfer to fetus. These changes include skeletal resorption and increased intestinal calcium absorption. Increased calcium requirement for milk production, hypoestrogenic state, prolactin-induced synthesis of parathormon related peptid (PTHrP) might also cause maternal bone resorption during breastfeeding period.[1,2] These changes are possible factors that facilitate the development of osteoporosis and fracture formation. Fortunately, this loss of bone mineral density (BMD) is temporary and bone density usually improves greatly within 6-12 months after breastfeeding ceases.[3] Nevertheless, the cause and etiopathogenesis of PLO have not been thoroughly elucidated.

CASE REPORT

A 28-year-old female patient applied to health center due to lower back and back pains that started one month after birth and spread to the left hip. It was her first pregnancy. The lumbar magnetic resonance imaging (MRI) was obtained and revealed fracture at L1 level and the patient was referred to our hospital. Thoracic MRI was performed because of an increase in severity of pain and newly developed fractures at L1, L2, T10, T12 vertebra levels were reported (Figure 1, Figure 2 A, B). On examination, the patient was uncomfortable. The temperature was 36.5°C, the pulse 84 beats per minute, the blood pressure 110/70

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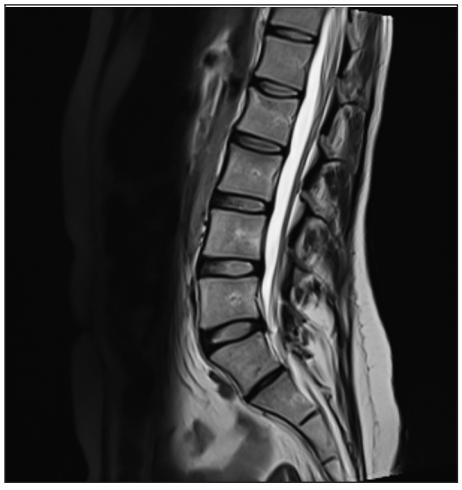

FIGURE 1: Sagittal T2WI shows the L1 and L2 vertebrae end plate fractures.

mm Hg. The abdomen was soft and non-tender, no peripheral edema was present. The back and low back region were tender on palpation.

BMD was performed. Lumbar spine and femoral neck Z -scores were L1:-3.0; L2: -3.6; L3:-2.5; L4:-2.6; femur neck:-2.5 respectively. It was stated that, menarche was at the age of 12 and she had regular menses. She reported no history of eating disorder or any bowel disease that cause malabsorption. There was no steroid use or smoking history. She was still breast-feeding when she applied. Menstruation began in the 3rd month after birth. Laboratory values were not remarkable except for low ferritin and vitamin D levels. Abdominal ultrasonography performed due to the lesion detected randomly in the liver during thoracic MRI and revealed an 8 mm stone in the left kidney. Secondary causes of osteoporosis were investigated and no cause was found. Laboratory test results are shown in Table 1.

The patient was informed about her process and her written approval was received for a case report presentation.

DISCUSSION

Pregnancy and lactation-related osteoporosis is a rare clinical condition which was first described by Nordin and Roper.[4] The incidence of PLO is reported to be 0.4 in 100 000 women.[5] Its etiology has not been fully explained. In addition to physiological processes that result in increased bone resorption during pregnancy and lactation, osteoporosis risk factors such as low calcium intake, vitamin D deficiency, physical inactivity, low BMI, genetic predisposition to osteoporosis and smoking are also considered to be risk factors for the development of PLO.[2,5] In order to ensure adequate calcium transfer to the fetus, 25-30 g of calcium transfer occurs from mother to fetus, 80% of which is in the last trimester.[6,7] For this purpose, maternal bone resorption and absorption of calcium from the intestines are increased. The increase in calcium absorption from the intestines is associated with increased production of 1.25 (OH) 2D3 in

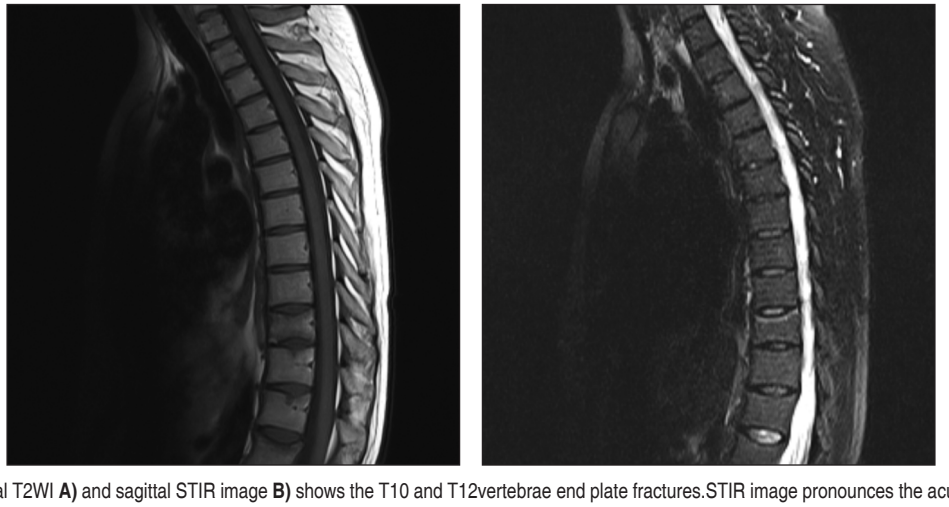

FIGURE 2: Sagittal T2WI **A)** and sagittal STIR image **B)** shows the T10 and T12vertebrae end plate fractures.STIR image pronounces the acute nature of fracture at T10 vertebrae end plate.

TABLE 1: Laboratory test results of the patient.

Tests	Values	Normal Range
25-OH Vitamin	7.7 L ug/L ↓	14-60
Calcium. total (Serum)	9.25 mg/dL	8.6-10
Phosphorus	3.3 mg/dl	2.5-4.5
Alkalen phosphatase (ALP)	99 U/L	25-100
Urinary calcium/creatinine ratio	0.08	<0.14
Parathormon. intact (PTH)	18.0 pg/mL	10-65
Calcitonin	4.4 pg/mL	0-18
Estradiol (E2)	143.00 pg/ml	11-44
HGB (Hemoglobin)	11.4 L g/dL ↓	11.9-14.6
HCT (Hematokrit)	37.5 %	36-44
Ferritin	10 ng/ml ↓	20-200
TSH	2.3 mIU / L	0.4 - 4

the placenta and kidney. Estrogen also increases calcium absorption from intestines during pregnancy.[6] It is suggested that PTHrP may be responsible for the increase of bone resorption occurring in the mother during pregnancy and low estradiol levels might contribute to this process. The PTHrP secreted from the placenta and breast reaches its highest level in the third trimester during pregnancy. PTHrP stimulates bone resorption via the PTH/PTHrP receptor.[3] Accordingly, the excretion of calcium in the urine increases.[8] In the ultrasonographic examination of our patient, stone formation was observed in the left kidney. The absence of a history of kidney stones suggests that the stone was formed due to hypercalciuria during pregnancy.

There are also physiological processes leading to osteoporosis during lactation. Studies have reported that bone mineral density decreases during lactation and is associated with high bone turnover.[9] It is suggested that the prolactin secreted during lactation reduces bone mineral density through parathormon-related peptid (PTHrP).[1,9]

These processes, which increase bone resorption and decrease BMD in the mother, are thought to predispose to vertebra fractures with mechanical load and postural changes due to pregnancy. Rarely, femur fractures can also be observed in PLO.[10] In our patient, Femoral neck Z score was -2.5 and it was osteoporotic.

As in many previously published PLO cases, the risk factors for osteoporosis was not observed in our patient except vitamin D deficiency. Although the need for calcium for milk production increases, the daily intake of calcium and vitamin D is recommended during pregnancy and lactation is the same as for women in the same age range.[11] In studies conducted, it is stated that calcium supplementation during lactation does not affect bone density of the mother.[12] Therefore, it may be thought that some risk factors that have not been revealed may play a role in the development of PLO.

While there are studies showing that BMD increases after pregnancy and lactation period without a specific treatment, some studies show an increased risk of fracture, especially in subsequent pregnancies.[13]

Weaning, calcium and vitamin D supplementation, anti-resorbtive and osteo-anabolic agent therapies are recommended in the treatment of the disease.[5] Although bisphosphonates have been shown to increase bone mineral density in PLO patients, their reliability has been questioned due to their long-term efficacy and accumulation properties.[5,14] Good results have been reported with the use of teriparatide and denosumab.[15] Our patient refused to stop breastfeeding and calcium and vitamin D treatment was started.

These patients generally present with lower back and back pains during the last trimester of pregnancy or during lactation. Since these pains are common during pregnancy, fractures due to pregnancy-related osteoporosis can be overlooked. Therefore, PLO should be considered in patients with persistent and severe pain and radiological evaluation should be performed. In all PLO cases, the causes of secondary osteoporosis should be investigated with detailed history and laboratory examination.

Source of Finance

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *F. Aytül Çakcı, Emel Bayrak;* ***Design:*** *F. Aytül Çakcı, Emel Bayrak, Barış Taşbaş;* ***Control/Supervision:*** *Emel Bayrak;* ***Data Collection and/or Processing:*** *Emel Bayrak;* ***Analysis and/or Interpretation:*** *Emel Bayrak, Barış Taşbaş;* ***Literature Review:*** *Emel Bayrak, Barış Taşbaş;* ***Writing the Article:*** *Emel Bayrak;* ***Critical Review:*** *F. Aytül Çakcı.*

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