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CASE REPORT

A case report: pregnancy-induced severe osteoporosis with eight vertebral fractures

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Abstract Osteoporosis associated with pregnancy and lactation is a rare condition. The prevalence, etiology and its pathogenesis is unknown. It causes one or more vertebral fractures with severe, prolonged back pain and height loss in affected women. Majority of the cases are seen in the third trimester or just after delivery in primagravid women. In this case report, a 30-year-old woman who had severe pregnancy-induced osteoporosis with 8 vertebral fractures was presented. During last month of her first pregnancy she had moderate back pain. After delivery, the back pain has gotten worse. The radiological examinations have shown that there was 50% in T_6, T_8 and T_{10}; 30% in L_2; 20% in L_1 height loss and biconcave vertebral images in $L_{3–5}$. In the bone mineral density, $L_{2–4}$ T score was −4.7 and total femoral T score was −3.1. There was no abnormality in the laboratory findings except mild elevation in alkaline phosphates. Although pregnancy-associated osteoporosis is a rare condition, when pain occurs in the last trimester or early postpartum period, it should be considered in differential diagnosis.

Introduction

Osteoporosis is a disease of aging which is commonly seen in patients after 50 years of age, although a number of young adults of both sexes have been affected without unidentifiable underlying cause [1]. Osteoporosis associated with pregnancy and lactation is a rare condition. The prevalence, etiology and pathogenesis of this osteoporosis are unknown. First description of pregnancy-induced osteoporosis was done by Albright and Reifenstein in 1948. Up to now approximately 100 cases were reported [2]. Majority of the cases are seen in the third trimester or just after delivery in primagravid women. It causes one or more vertebral fractures with severe and prolonged back pain and height loss in the affected women. This case report indicated a 30-year-old woman who had severe pregnancy-induced osteoporosis with 8 vertebral fractures. In the literature, there was not any pregnancy-associated osteoporotic case with more than five vertebral fractures according to our knowledge. The informed consent was obtained from the subject.

Case

A 30-year-old woman during the last month of her first pregnancy had moderate back pain. After delivery, her back pain has gotten worse. She was previously diagnosed as myofacial pain syndrome in other medical center and treated with physical modalities (heat application and TENS) and analgesic-myorelaxants. These treatment procedures did not help her pain. Therefore, at 3 months following delivery, she was admitted to the physician with complains about severe back pain and height loss. She was not able to carry and breast-feed her baby due to severe pain. She could breast-feed her baby only 1 month because of using pain killer. She had difficulties with her daily activities as well. There were tenderness in thoracic vertebrae and spasm on vertebral muscles. Even though the spinal

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range of motion was limited, there was no abnormal neurological sign in physical examination. The pain was localized to back and there was no numbness and/or weakness on lower limbs. She had apparent thoracic kyphosis. Her height was 152 cm, arm span was 164 cm and weight was 50 kg. She reported that her previous height was 161 cm indicating 9 cm height loss.

Radiological examination

On lateral spinal radiographs there was variable degrees height loss (50% in T_6, T_8 and T_{10}, 30% in L_2 and 20% in L_1) in thoracolomber vertebral bodies and biconcave vertebral images through $L_{3–5}$ (Fig. 1). There was also mild scoliosis on antero-posterior view of spinal X-ray (Fig. 2). These results are consistent with the magnetic resonance imaging and bone scan (Figs. 3, 4).

Bone mineral density

Bone mineral density was measured by using a dual-energy X-ray absorptiometry (Lunar, Madison) and the results showed that $L_{2–4}$ T score was −4.7 and total femoral T score was −3.1 that indicated severe bone loss.

Biochemical evaluation

The laboratory assessments including erythrocytes sedimentation rate (ESR), calcium (Ca), phosphorus (P), creatinine (Cr), liver enzymes (AST, ALT), alkaline phosphates (ALP), thyroid hormone (TSH), para-thyroid hormone (PTH), osteocalcin, 25-hydroxyvitamin D (25-OHD) and urinary deoxypyridinoline revealed no abnormality except a mild elevation in ALP. The protein electrophoresis was in normal range as well.

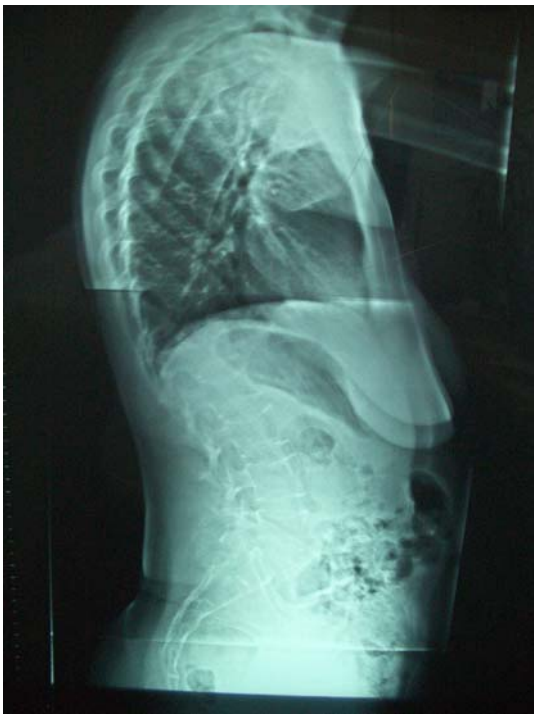

Fig. 1 Lateral spinal X-ray shows thoracal kyphosis

Fig. 2 Antero-posterior spinal X-ray

She reported that she was completely healthy before pregnancy. She did not have any disease causing osteoporosis and did not use any medicine such as corticosteroid or thyroid hormone. She was a non-smoker and there was no positive family history for osteoporosis or related risk factors. However, it was noted that she did not take calcium supplementation during her pregnancy. There were no signs of metabolic, metastatic or infectious bone disease based on laboratory and radiological findings. According to these findings she was diagnosed as pregnancy-induced osteoporosis. Alendronate 70 mg/week, elementary calcium 1,000 mg/day and vitamin D3 400 IU/day were started immediately. A thoracolumbosacral orthosis (TLSO) was prescribed. In addition, she was advised to bottle-feed her baby. Analgesics were recommended for pain management.

The back pain has gradually decreased within 3 months, although it continued as long as about 6 months. Using TLSO was discontinued 3 months after. A rehabilitation program including muscle strengthening, range of motion and relaxation exercises as well as weight-bearing exercises was started.

At an 1-year follow-up, she did not have any complain about back pain. She felt better than before and was

Fig. 3 Sagital T_1-weighted MRI of thoracolomber spine demonstrates multiple osteoporotic vertebral fractures and codfish appearence

Fig. 4 Tc-99 bone scan shows increased uptake in affected vertebrae

comfortable with her daily activities. There was no new vertebral fracture or progressive collapse on the already existed vertebrae. The T scores increased to −3.2 in $L_{2–4}$ and −2.8 in total femoral region. Currently, she is under antiosteoporosis treatment (alendronate 70 mg/week, calcium 1,000 mg/day and vitamin D3 400 IU/day) and continues the rehabilitation program.

Discussion

Pregnancy-induced osteoporosis is a rare problem of unknown cause. It is frequently seen in the last trimester or early postpartum period. Why some women tend to develop pregnancy-induced osteoporosis has not been understood. A positive family history of postmenopausal osteoporosis in pregnancy-induced osteoporosis is seen more frequently than the controls. It is thought to suggest that some hereditary factors are involved [3]. Ghannam et al. [4] stated that the number of pregnancies and total duration of lactation negatively correlated with BMD Z score at the spine and Saudi women have significantly lower BMD levels than USA counterparts. They concluded that this difference

might be caused by increased number of pregnancies and longer duration of lactation together with prevalent vitamin D deficiency. However, reproduction-induced bone loss are apparently multifactorial phenomena that may be related not only to duration and magnitude of lactation and/or duration of postpartum amenorrhea but also to dietary factors and other yet unknown individually modulated factors [5]. Dynamic changes in bone and calcium metabolism in women may also contribute pregnancy or lactation-induced osteoporosis. During pregnancy, especially in the third trimester, considerable amounts of calcium are transported from the mother to fetus through the placenta for normal bone mineralization in the growing fetus. However, in nursing mother calcium losses are greater than those in pregnant woman. Lactating woman provide an average of 200–250 mg calcium/day to their infants, sometimes as much as 400 mg/day. Therefore, maternal metabolism of bone and calcium changes dynamically during pregnancy and lactation [6]. In our case, she did not receive calcium supplementation during pregnancy and after delivery till diagnosis was done. So, we suggest that her osteoporosis may result from negative calcium balance during the last trimester and early postpartum period.

O'Sullivan et al. [2] retrospectively investigated 11 pregnancy and lactation-associated osteoporotic women. Ten of their 11 cases had more than 1 fragility vertebral fractures. However, the most number of vertebral fractures was 4 in 4 of 10 cases in their study. Our case has eight vertebral fractures showing severity of the disease. To the best of our knowledge this is the first case in the literature that has more than four vertebral fractures due to pregnancy-associated osteoporosis without trauma. In O'Sullivan et al. study, 9 of 11 patients received biphosphanate treatment for a median duration of 24 months. After 2 years of the biphosphanate therapy, the spinal bone density increased by 23% over baseline value. Our case has taken alendronate for 1 year and the improvement of the bone density was about 32%.

Generally, spinal BMD value in patients with pregnancy-associated osteoporosis is lower than those in femoral site [2, 7]. In O'Sullivan's study, the mean T score of lumbar region was −2.8 while that was −1.9 in the femoral region [2]. In another study, this was −3.34 and −2.41 in both the lumbar and femoral regions, respectively [7]. This result is similar with our result. The lumbar BMD (T score: −4.7) was lower than those in femoral region (T score: −3.1) in our case as well.

Clinical feature of pregnancy-induced osteoporosis is similar with the other type of osteoporosis. The most common symptom is severe back pain in the lower thoracic and/or lumbar region [8]. In some patients back pain may resolve spontaneously within month(s), although others have prolonged and severe back pain. In our case, back pain was in both thoracic and lumbar spinal region and very severe. She was not able to carry her baby and had limitation in her daily living activity because of back pain. The back pain was gradually decreased; however, it lasts at 6 months of follow-up period. During this time she had to use analgesics -if needed- for her severe back pain.

Antiresorptive agents considered first-line treatment for osteoporosis [9]. Even though, long-standing effects of biphosphonates are not known well, for the last 10 years they have also been used pediatric patients with osteoporosis [10]. In this case, because of very severe osteoporosis, alendronate and Ca plus Vitamin D were used for the treatment after the delivery and, while using these drugs, the breastfeeding has been stopped.

Another medical treatment option could be teriparatide which is a bone formation agent that has been shown to increase bone mineral density and reduce the risk of fractures [11]. It is indicated for the treatment of postmenopausal women with osteoporosis who are at a high risk for fracture or for women who have failed or are intolerant of previous osteoporosis treatment therapy [12]. Also, most of teriparatide user were older, more severely ill, and appeared to have more severe osteoporosis than patients using antiresorptive [9]. The case was not in the postmenopausal period and she was relatively young, teriparatide was not used as being the first-line treatment. Moreover, we could not find about teriparatide treatment in a young woman with pregnancy-induced osteoporosis, it can be chosen for a treatment agent. However, during the treatment, intolerance of the antiresorptive treatment was not observed and the treatment regiments were not failed, therefore, current antiresorptive therapy was continued.

Other treatment modalities may be indicated such as kyphoplasty or vertebroplasty in patients who are resistive to medical treatment. These percutaneous interventions are very efficient for pain relief in acute fractures. MRI is very useful tool to distinguish acute or subacute fractures from healed fractures. The finding of bone marrow edema on MRI (hypointense signal on T1 and hyperintense signal on T2) is extremely helpful in predicting which patients with osteoporotic fractures are more likely to respond to kyphoplasty. Although some height restoration of collapsed vertebra may be obtained with this technique, the effect of kyphoplasty to prevent deformity is limited especially for late fractures [13]. Bayram et al. [14], presented a 37-year-old woman with spinal vertebral fractures due to pregnancy-associated osteoporosis who was treated with antiosteoporotic drugs (alendronate, calcitonin, calcium and vitamin D) and vertebral kyphoplasty for three vertebral level (L_{2-4}). They reported good pain relief, however, 2 weeks after the kyphoplasty 2 new vertebral fractures localized at L_1 and T_{11} occurred. After new fractures they advised to their patient to wear a thoracolumbosacral orthosis, limit physical activities and continue the same medication.

We conclude that if a pregnant woman or woman in postpartum period has persistent back pain, pregnancy and lactation-induced osteoporosis should be kept in mind in the differential diagnosis. This rare type of osteoporosis is associated with fragility fractures resulting disabilities and difficulty in daily activities and even breast-feeding. Therefore, it should be managed properly as early as possible when it is diagnosed.

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