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CASE REPORT / ПРИКАЗ БОЛЕСНИКА

# Pregnancy- and lactation-associated osteoporosis with vertebral fractures

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**SUMMARY**

**Introduction** Pregnancy- and lactation- associated osteoporosis (PLO) is a rare disease for which the pathophysiological mechanism is as yet incompletely known. The incidence of PLO is 0.4 in 100,000 women. It is considered that the number of undiagnosed patients is even higher. PLO can lead to multiple fragility compression fractures in the spinal vertebrae.

**Case outline** We present the case of a 30 years old woman (first-born, breastfeeding child) who came for examination due to lower back pain that occured after childbirth without any apparent cause. The patient was found to have low levels of vitamin D and low bone mineral density on osteodensitometry (established osteoporosis). Magnetic resonance imaging (MRI) examination showed vertebral bodies fractures Th11, Th12 and L4. During therapy, we used vitamin D (800 IU/24 h), alendronate (70 mg once weekly), calcium 1000 mg/24h and thoracic lumbar sacral orthosis (TLSO) as support to spine. After 12 months of treatment osteodensitometry findings were close to normal, control MRI showed no further collapse of vertebral bodies and clinical examination of spine was orderly.

**Conclusion** PLO is a rare clinical condition and it must be kept in mind in the differential diagnosis in patients having low back pain during or after pregnancy. Early diagnosis and treatment of PLO and regular follow-up of these cases are particularly important. The the stability of the spine in patients with vertebral fractures must be carefully monitored as well as using the TLSO as a support for the spine.

**Keywords:** osteoporosis; pregnancy; lactation; vertebral fractures

## INTRODUCTION

Osteoporosis is the most common bone disease in humans. It is characterized by low bone mass, deterioration of bone tissue and disruption of bone architecture, compromised bone strength and an increase in the risk of fracture. Pregnancy- and lactation-associated osteoporosis (PLO) is a rare disease for which the pathophysiological mechanism is as yet incompletely known [1]. The incidence of PLO is 0.4 in 100,000 women. Approximately 100 cases of pregnancy and lactation-associated osteoporosis were described between 1955. and 2009 [2]. It is considered that the number of undiagnosed patients is even higher. PLO led to multiple fragility compression fractures in the spinal vertebrae [3]. Analysis of diagnosis and treatment of this case can help clinicians to pay attention to the disease and the evaluation of osteoporosis. The clinical signs usually become to manifest in the postpartum period when patients come for examination with more pronounced back pain.

## CASE REPORT

A 30-year-old patient (breastfeeding a firstborn child) came in for examination due to lower back pain that occurred after childbirth without any apparent cause. She breastfed for a month and menstruation has returned to normal. She went to a local clinic for treatment, but the pain did not lessen. Her medical history was not remarkable for chronic disease, drug use, smoking or alcohol use. The examination was done one month after the delivery. Examination revealed pain in the lower back and weaker movements in the spine. The performed laboratory findings were shown in Table 1. Ultrasound examination of the abdomen was within physiological limits. The gynecological examination was orderly. Neurological examination showed no signs of neurological diseases. Urological examination showed no signs of urological diseases or problems. The patient was referred for magnetic resonance imaging (MRI) of the spine, which showed compressive fractures of vertebral bodies Th11, Th12 and L4, Figure 1.

Considering the reduced values of vitamin D and the findings of MRI she was referred for

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**Table 1.** Laboratory findings at the first examination

| Blood work | Value | Units | Reference values |
|---|---|---|---|
| WBC | 3.7 | $10^9$/l | 3.9–10 |
| RBC | 4.67 | $10^{12}$/l | 3.9–5.9 |
| Hgb | 130 | g/l | 110–180 |
| Plt | 190 | $10^9$/l | 140–450 |
| CRP | 2 | mg/l | < 5 |
| Urea | 4.4 | mmol/l | 1.6–7.5 |
| Creatinine | 57 | umol/l | 53–124 |
| Ca (total) | 2.36 | mmol/l | 2.02–2.60 |
| Inorganic P | 3.68 | mmol/l | 0.81–1.62 |
| Vitamin D | 40.3 | nmol/l | 75–250 |
| T3 | 1.43 | ng/ml | 0.79–1.58 |
| T4 | 9.18 | µg/dl | 4.9–11 |
| TSH | 0.791 | IU/ml | 0.27–4.2 (adults) |
| PTH | 66.3 | pg/ml | 15–66 |

WBC – white blood cells; RBC – red blood cells; Hgb – hemoglobin; Plt – platelets; CRP – C-reactive protein; T3 – triiodothyronine; T4 – thyroxine; TSH – thyroid-stimulating hormone; PTH – parathyroid hormone

**Table 2.** Osteodensitometry test findings during the treatment

| Osteodensitometry | First examination | After five months | After 12 months |
|---|---|---|---|
| L1 (BMD g/cm$^2$) | 0.87 | 0.787 | 0.987 |
| L2 (BMD g/cm$^2$) | 0.83 | 0.714 | 1.143 |
| L3 (BMD g/cm$^2$) | 0.75 | 0.783 | 1 |
| L4 (BMD g/cm$^2$) | 0.62 | 0.754 | 0.934 |
| Total (lumbar) (BMD g/cm$^2$) | 0.744 | 0.766 | 1.014 |
| T score (lumbar) | -2.8 | -2.4 | 0.4 |
| Z score (lumbar) | -2.8 | -2.4 | -1.9 |
| Neck (BMD g/cm$^2$) | 0.571 | 0.61 | 0.98 |
| Total neck (BMD g/cm$^2$) | 0.642 | 0.668 | 0.98 |
| T score (femur) | -2.2 | -2.2 | 1 |
| Z score (femur) | -2.2 | -2.2 | 1 |

BMD – bone mineral density

**Table 3.** Values of vitamin D in blood during the treatment

| Vitamin D (nmol/l) | First examination | After three months | After five months | After 10 months | After 12 months |
|---|---|---|---|---|---|
| | 40.3 | 54 | 72 | > 75 | > 75 |

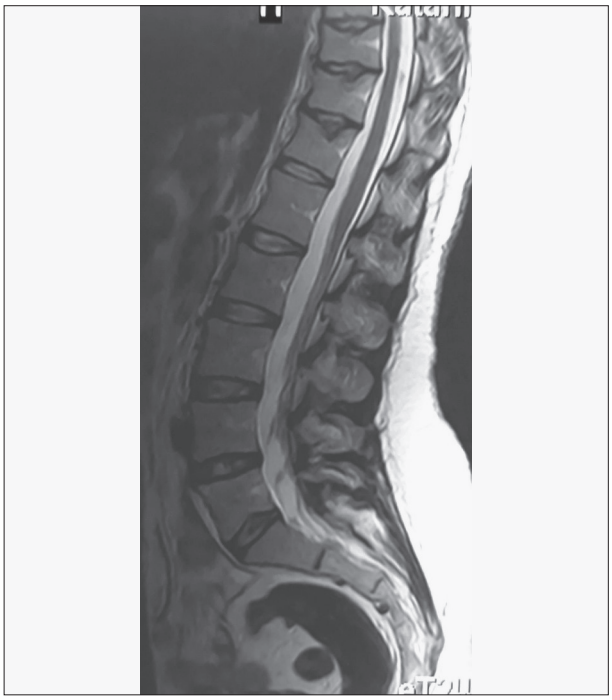

**Figure 1.** Magnetic resonance imaging of thoracolumbar spine; compressive fractures of the vertebral bodies Th11, Th12, and L4

osteodensitometry test, which confirmed the diagnosis of osteoporosis, Table 2.

Bisphosphonates (alendronate 70 mg once weekly), vitamin D (800 IU/day), calcium (1200 mg/day), analgesics and thoracic lumbar sacral orthosis (TLSO) were prescribed in the therapy and advice was given to stop lactation. The level of vitamin D in the blood was monitored.

After five months, the osteodensitometry test values, although better, still did not satisfy nor did the analysis of vitamin D levels (72 nmol/l), Table 3.

Bisphosphonates and vitamin D were continued in the therapy. The pain in the spinal column stopped, and the

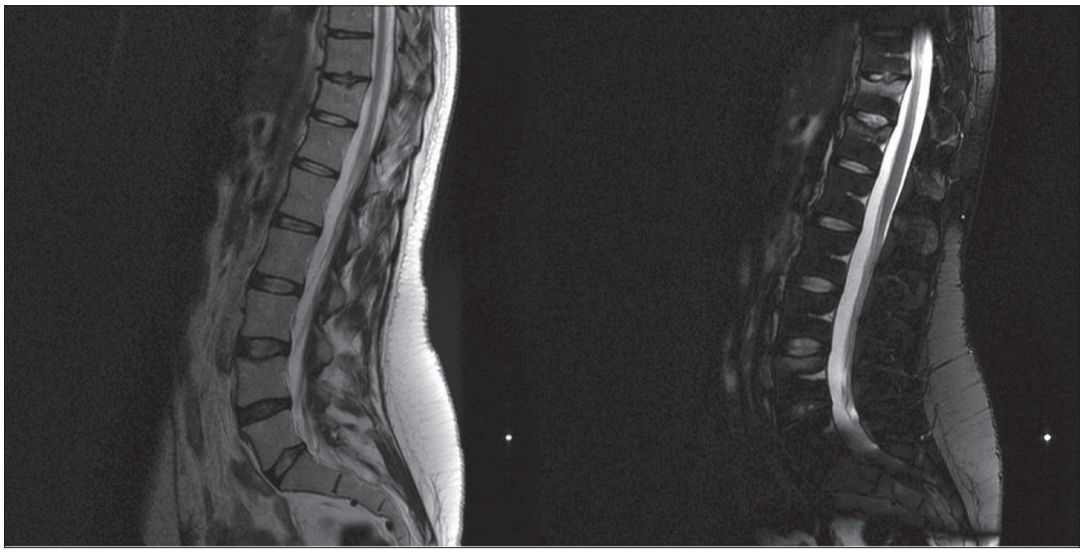

**Figure 2.** Control magnetic resonance imaging of the thoracolumbar spine after 10 months

movements of the spinal column were normal. Wearing TLSO continued for four hours a day.

Ten and 12 months after the first examination, the control values of vitamin D were normal, and 12 months after first examination the osteodensitometry values were close to normal. The patient had orderly clinical findings. 12 months after initial treatment laboratory findings of bone metabolism are normal and we stopped bisphosphonates and advised the patient to check blood levels of vitamin D every three months. Control MRI after 12 months did not show further vertebral collapse, Figure 2.

Written consent for publication of this article has been obtained by patient.

## DISCUSSION

PLO is a condition that is often overlooked, although it was described as early as 1955 [4]. The causes of PLO are still unclear. There are risk factors for PLO development such as smoking, malnutrition, lower calcium and Vitamin D intake, poor mobility, weight gain during the last trimester and oligomenorrhea [5]. Cohen et al. [6] considered that women with PLO may have a low bone remodeling state assessed more than a year postpartum, which increases understanding of the pathogenic mechanism of PLO and also may have underlying osteoblast functional deficits which could affect their therapeutic response to osteoanabolic medications. In our case, there were no risk factors.

Careful analysis of the examination of patients with suspected PLO is required. A comprehensive analysis of other conditions that would lead to osteoporosis is also needed. A detailed evaluation of bone metabolism is extremely important [7]. Our patient was a young woman with normal period who suffered with unexplained pain, has brittle fractures and decreased bone mineral density (BMD). In this case, we first took into consideration the metabolic bone diseases, which had to be carefully identified with osteoporosis and other metabolic bone diseases. In our case we performed complete laboratory findings of bone metabolism, which only showed a reduced level of vitamin D, so we raised the level of vitamin D to normal levels, along with calcium intake.

Bisphosphonates can be used regularly in PLO therapy, and there was increase in BMD after bisphosphonate use in patients with PLO [8, 9, 10]. For increasing BMD can be used teriparatide [11] and also alfacalcidol can be used for PLO joined with vertebral fractures [12]. Osteodensitometry examination of our patient showed a strong decrease in BMD and Z-score, so we decided to use bisphosphonates in therapy. We used bisphosphonates in therapy for 12 months.

Strong loss of bone mass in the vertebral bodies can cause collapse and compressive fractures with strong back pain [13]. Vertebral fractures occur most commonly in PLO and are often multiple [14]. If a vertebral collapse is found, TLSO should be used. Sometimes the stability of the spine is disturbed, so surgical treatment with kyphoplasty is necessary [15]. The stability of the spinal column should be constantly monitored by control MRI. In our case, there was an affection of the vertebral bodies of the Th11, Th12, and L4. With rapid diagnosis and timely therapy for further progression of osteoporosis, we used TLSO for treatment and prevented further vertebral collapse. Our last MRI examination at the end of treatment did not show further collapse of the vertebral bodies. The patient's clinical condition was normal after 11 months.

Breastfeeding produces an obligatory loss of maternal skeletal mineral which contributes to the decline of bone density [16]. All women with developed PLO are advised to stop breastfeeding.

PLO is a rare clinical condition and it must be considered in the differential diagnosis in patients coming with low back pain during or after pregnancy. Although not specified as a diagnostic criterion, the exclusion of other reasons for osteoporosis and progressive clinical course are necessary and helpful in the diagnostic process. At the same time, the monitoring and evaluation of the efficacy of PLO intervention is also very important. The fractures of vertebral bodies related to PLO may be an important cause of disability in the long term. Early diagnosis and treatment of PLO and regular follow-up of these cases are particularly important. The stability of the spine in patients with vertebral fractures must be carefully monitored and the TLSO must be used to support the spine.

**Conflict of interest:** None declared.

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## Остеопороза током трудноће и лактације са преломима кичмених пршљенова

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**САЖЕТАК**

**Увод** Остеопороза изазвана трудноћом и дојењем је ретка болест за коју је патофизиолошки механизам још увек непознат. Њена инциденца је 0,4 на 100.000 жена. Сматра се да је број недијагностикованих болесница још већи. Може изазвати компресивне преломе пршљенова.

**Приказ болесника** Представљамо случај 30-годишњакиње (прворотка, дојиља) која је дошла на преглед због болова у доњем делу леђа, који се јављају после порођаја без икаквог очигледног разлога. Утврђено је да болесница има нижи ниво витамина *D* и ниску густину костију на остеоденситометријском прегледу (утврђена остеопороза). Преглед магнетном резонанцом показао је преломе тела пршљенова *Th*11, *Th*12 и *L*4. У терапији смо користили витамин *D* (800 ИЈ / 24 ч.), алендронат (70 мг једном недељно), калцијум 1000 мг / 24 ч. и спиналну ортозу као потпору кичми. После 12 месеци лечења остеоденситометријски налази су били близу нормале, а контролни преглед магнетном резонанцом није показао даљи колапс тела пршљенова. Клинички преглед кичме је био уредан.

**Закључак** Остеопороза изазвана трудноћом и дојењем је ретко клиничко стање и мора се имати на уму у диференцијалној дијагнози код болесница које имају болове у леђима током или после трудноће. Рано дијагностиковање, лечење и редовно праћење случајева остеопорозе изазване трудноћом и дојењем посебно су важни. Стабилност кичменог стуба код болесница са преломима пршљенова мора се пажљиво проценити и надгледати, и мора се користити спинална ортоза као потпора кичми.

**Кључне речи:** остеопороза; трудноћа; лактација; преломи пршљенова