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# Pregnancy associated osteoporosis – a case report

## Osteoporoza związana z ciążą – opis przypadku

Dytfeld Joanna, Horst-Sikorska Wanda

Katedra i Zakład Medycyny Rodzinnej,
Uniwersytet Medyczny im. Karola Marcinkowskiego w Poznaniu, Polska

### Abstract

*Loss of bone mineral density (BMD) – usually temporary – occurs during pregnancy and lactation. Pregnancy associated osteoporosis (PAO) is an uncommon disease of unknown etiology. We present a case of a 35-year old woman with PAO, manifesting initially at the end of the first pregnancy as back pain. It reappeared in the second pregnancy, four years later. X-ray revealed multilevel compression fractures of Th12, L1, L2. DEXA showed L2-L4 T-score: -3.3 SD, hip T-score: -2.09 SD. Laboratory findings were irrelevant. She was put on antiresorptive treatment, calcium and vitamin D. Although there has been an improvement in BMD, the patient is a definite candidate for vertebral kyphoplasty due to disabling pain.*

Key words: **pregnancy** / **lactation** / **osteoporosis** / **bone mineral density** /

### Streszczenie

*W czasie ciąży i laktacji dochodzi do utraty gęstości masy kostnej, jednak najczęściej jest ona przejściowa. Osteoporoza związana z ciążą (pregnancy associated osteoporosis – PAO) jest rzadką chorobą o nieznanej etiologii. Prezentujemy przypadek 35-letniej kobiety, u której choroba manifestowała się początkowo pod koniec pierwszej ciąży bólem pleców. Pojawiły się one także w drugiej ciąży, cztery lata później. Rtg ujawniło wielopoziomowe złamania kompresyjne Th12, L1, L2. Densytometria wykazała T- score L2-L4: -3,3 SD, T-score z bliższego końca kości udowej: -2,09 SD. Wyniki badań laboratoryjnych nie wykazały istotnych patologii. Chora została poddana leczeniu przeciwresorpcyjnemu, preparatami wapnia i witaminy D. Mimo, że nastąpiła poprawa w zakresie parametrów densytometrycznych, ból w dalszym ciągu znacznie upośledza życie pacjentki i powoduje, że jest ona kandydatką do vertebroplastyki.*

Słowa kluczowe: **ciąża** / **laktacja** / **osteoporoza** / **gęstość mineralna kości** /

**Corresponding author:**
Joanna Dytfeld
Katedra i Zakład Medycyny Rodzinnej
Uniwersytet Medyczny im. Karola Marcinkowskiego
Ul. Przybyszewskiego 49, 60-355 Poznań
tel. 061 8691147, 606 833132, fax. 061 8691143
e-mail: dytfeld@poczta.onet.pl

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## Introduction

Osteoporosis is a disease of an old age, most prevalent in postmenopausal women. Pregnancy associated osteoporosis (PAO) is a rare condition of unknown etiology. Since the first report in 1948, about 100 cases have been described [1]. It manifests generally in primiparas at the end of pregnancy or in the postpartum period and presents with rapid onset of severe back pain. It is often a result of multiple vertebral fractures, that may be an indication for surgery in the postpartum period. Rarity of the disease often causes a significant delay in diagnosis. Thus, patients might not be treated accurately and may suffer from life-threatening complications. In this paper we present a case of a 35-year old woman with a long history of osteoporosis diagnosed during the second pregnancy.

## Case presentation

A 35-year-old Caucasian female was consulted at the Endocrinology Outpatient Clinic, University Hospital No. 2 in Poznan, Poland, due to growing intensity of back pain. Complaints began in the last trimester of the first pregnancy (the patient was then 22 years old) and were classified as non-specific low back pain, with no further diagnostics. The patient was breastfeeding for the next three months. In 2001, at the end of the third trimester of her second pregnancy, girdle pain in the thoracic and lumbar spine substantially intensified. Neurological examination was unremarkable with no focal signs. The pregnancy ended in cesarean section (healthy male, birthweight 3250 g). The woman was hospitalized at the Department of Neurology. The X-ray showed multilevel fractures of Th12, L1, L2. Osteoporosis complicated by compression vertebral fractures was diagnosed. Laboratory tests revealed serum levels of calcium, alkaline phosphatase and parathyroid hormone to be normal, phosphate excretion in the urine was slightly reduced.

In 2001, patient's height was 162 cm (2-cm defect in comparison to the pre-pregnancy height). Subsequently, she was discouraged from lactation, provided with the Javette's corset, treated with salmon calcitonin nasal, alfacalcidol and NSAIDs, which relieved the pain. Diagnostic test for malabsorption syndrome was also undertaken but it was negative. DEXA (dual X-ray absorptiometr) (Lunar DPX-L, Lunar Inc., Madison, Wi, USA) showed the following parameters of bone mineral density (BMD):

- L2-L4 BMD: 0.804g/cm², T-score: -3.3 SD, Z-score: -3.17 SD
- hip BMD: 0.729g/cm², T-score: -2.09 SD, Z-score: -2.06 SD
- distal forearm BMD: 0.250g/cm², T-score: -3.48 SD, Z-score: -3.48 SD.

MRI of the spine performed 3 months after the delivery confirmed compression fracture of Th8 and L1 and infraction of upper epiphyseal plate of 10th, 11th and 12th thoracic vertebrae. Neither neoplastic invasion nor compression of the spinal cord were found. The patient was recommended therapy with antifracture medications accompanied by supplementation of calcium and vitamin D, which were continued in the next years (salmon calcitonin nasal, alendronate sodium, risedronate sodium).

The patient, gravida 2, para 2, no miscarriages, had her first menarche and has been menstruating regularly.

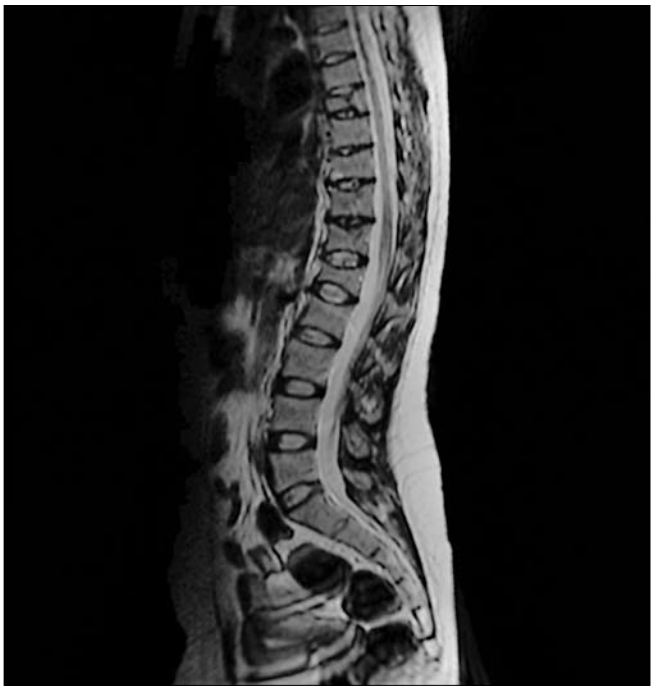

**Figure 1.** MRI of the thoraco-lumbar spine. Codfish appearance of Th8 and L1. Discrete bulging of fibrous rings.

During the pregnancies there was no calcium supplementation. Before the diagnosis of osteoporosis the patient had not suffered from any serious diseases, and had not been diagnosed with any chronic diseases. She also had not taken any drugs that might have affected bone metabolism, including hormonal drugs. Her calcium intake was estimated at 600mg per day. There were no low-energy fractures in her family history. For three years before the first pregnancy the patient smoked 5 cigarettes/day

Physical examination in 2011 revealed – height 161 cm, weight 62 kg; pain on palpation and limitation of movement of thoracic and lumbar spine, painful tension of paraspinal muscles. No abnormalities were found on neurological examination. Medication taken a the time were: risedronate 35 mg/week, alfacalcidol 1 mg/day, calcium carbonate 400mg/day. Periodically, the pain has been intensifying and remarkably limiting the mobility of the patient. Since the diagnosis she has been receiving a disability pension. Attempts to take up even part-time job failed due to pain, increasing over the last three months preceeding the visit. Spine X-rays confirmed previous findings and revealed no new ones. Because back pain was poorly responsive to treatment, the patient was referred to MRI, (Figure 1), which showed the deepening of the vertebral upper and lower lamina from Th8 to L1. The greatest changes occurred in the vertebral bodies of Th8 and L1, where codfish appearance was seen. Intrathecal movements were not demonstrated. Adjacent intervertebral discs revealed signs of degenerative changes - reduction in height (except for Th12/L1 and L1/L2), presence of small gas bubbles (vacuum symptom) and discrete bulging of fibrous rings. Slight compression of the anterior part of the dural sack was seen. Visualized spinal cord and cauda equina roots were normal.

Current BMD: L2-L4 - 0.971 g/cm², T-score: -1.91 SD, Z-score: -1.77 SD.


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Continuation of the treatment and rehabilitation program were advised.

## Discussion

Osteoporosis is a condition characterized by low bone mineral density and disturbed bone microarchitecture, which in turn leads to fractures. In postmenopausal osteoporosis there are certain risk factors for fracture, which can be identified and even quantified. On the other hand, etiopathogenesis of PAO remains unclear. So far, it has not been clearly explained why some pregnant women suffer from severe loss of bone mass, whereas others do not. In the presented patient the classical risk factors for osteoporosis and fracture were not present. However, there are data reporting they do co-exist in women with PAO more often than in healthy ones (for example low-energy fracture in family history) [1].

The fact that raises interest in this particular case is the relatively late menarche - in Poland the average age of menarche is 12 years and 9 months [2]. One cannot exclude that deterioration of bone quality might have occurred in the first pregnancy, as it is usually reported in PAO, but was not diagnosed then. Similar situation was described in the literature [3]. In addition, in women with previously diagnosed PAO, fractures in subsequent pregnancies might happen, although it is not common [1]. Patients should be informed about such risk.

Parity and lactation – contrary to popular opinions – do not contribute to permanent deterioration of BMD. The fact remains that there are changes in the calcium metabolism during pregnancy – the fetus receives an amount of calcium representing only 2-3% of its total content in the mother's body. Even despite this, the mother adapts to the changing conditions through increased intestinal absorption and renal reabsorption of calcium and vitamin D synthesis in the kidneys [4]. Undeniably, the average loss of BMD during pregnancy is ca. 5%, however, current data show that after weaning BMD returns to baseline within 6-12 months [5]. Such condition corresponds with the concept of transient osteoporosis of pregnancy (TOP) [6].

Moreover, most of the studies report lack of difference in both BMD and incidence of osteoporosis in later life among women who were pregnant at least once and nulliparas [7]. The former were shown to have 3-5% higher BMD than the latter [8]. Large clinical studies – SOF [9], MEDOS [10], EVOS [11] - reported the same incidence of fractures in women who gave birth and nulliparas.

There are several hypotheses attempting to explain the etiopathogenesis of PAO. The main reason for low BMD is most probably the loss of the protective effects of estrogens on bone and hyperprolactinemia accompanying lactation. It is also postulated that bone resorption in the course of PAO may be the result of increased secretion of PTHrP from the mammary gland during lactation [12]. There are isolated reports of calcitonin deficiency adversely affecting bone [5]. Some authors have also raised the issue of pre-pregnancy osteopenia, which is difficult to verify due to paucity of data on PAO.

Women with PAO were usually shown to have lower BMD and T-score in the lumbar spine than in the hip [13]. We confirmed this observation (T-score = -3.3 SD vs. -2.0 SD, respectively).

In addition to kypho- and vertebroplasty in the postpartum period that followed vertebral fractures in course of PAO and were presented elsewhere, low-energy fractures of acetabulum and femoral neck which required total hip replacement were also reported [14].

Importantly, there is a question of the recognition of PAO, which is de facto premenopausal osteoporosis. WHO definition of osteoporosis is based on BMD and T-score by means of DEXA. However, it applies to postmenopausal osteoporosis and therefore should not be used to diagnose younger patients. In such cases the International Society for Clinical Densitometry recommends to use DEXA Z-score. A Z-score of -2.0 or lower is defined as 'below the expected range for age', and a Z-score above -2.0 is 'within the expected range for age'. The diagnosis of osteoporosis was undeniable in the case of our patient, not only because of the densitometric parameters but also due to low-energy fractures.

Because of the rarity of PAO and its unknown etiology, there are no established treatment protocols and long-term effects of such therapy. Women usually benefit from treatment with bisphosphonates and calcitonine. There were also some reports about cases of strontium ranelate therapy [1]. Some authors reported that in women with PAO treated with bisphosphonates, the increase in BMD is relatively higher than in patients with postmenopausal osteoporosis [15].

In conclusion, it should be clearly stated that special attention should be given to pregnant women with back pain, that can be indicative of PAO – a rare, but serious disease with significant clinical implications.

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