

# Multiple Vertebral Compression Fractures Secondary to Pregnancy-induced Osteoporosis: A Case Report

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## Learning Point of the Article:

One must always consider the significance of sagittal imbalance and the potential role for operative orthopedic intervention or even correction, particularly when managing such a young patient cohort with such a severely morbid disease process such as osteoporosis.

## Abstract

**Introduction:** Although rare in incidence, pregnancy-induced osteoporosis (PIO)-associated OVCFs represent a significant cause of morbidity for the young, peri-partum female population.

**Case Report:** We present the case of a 27-year-old nulliparous lady who suffered seven osteoporosis vertebral compression fractures (OVCFs) with associated sagittal imbalance, the challenges posed to the attending physician or surgeon in treating this rare condition, as well as an in-depth discussion of previous literature reported on pregnancy-induced osteoporosis (PLIO) to date. Although rare in incidence, PLIO-associated OVCFs represent a significant cause of morbidity for the young, peripartum female.

**Conclusion:** This case demonstrates how multiple PLIO-associated OVCFs may be managed successfully, with careful consideration of sagittal imbalance, using a combination of medical and non-operative orthopedic therapies at medium-term follow-up.

**Keywords:** Pregnancy, Osteoporosis, Vertebral Compression Fractures, PLIO, Management

## Introduction

While osteoporosis is a common metabolic bone disease which occurs in both genders, its incidence tends to be much higher in the female population, particularly in the postmenopausal cohort [1]. Although the vast majority of female cases of osteoporosis focus on the postmenopausal patient, recent literature has demonstrated that peripartum females can be susceptible to pregnancy-induced osteoporosis (PIO) [2]. More commonly, potential breastfeeding-associated osteoporosis is widely acknowledged in the literature; however, its effect on actual bone loss has been deemed transitionary, with many physiological and homeostatic mechanisms at play postpartum [3].

A full-term neonate requires approximately thirty grams of calcium during pregnancy, with the majority of calcium transfer from mother to fetus occurring in the third trimester [4]. Although pregnancy has been described as a physiologic absorptive hypercalciuric state, rarely, more severe changes with decreases in bone mineral density have been described [5]. PIO conditions, such as post-pregnancy spinal osteoporosis and transient osteoporosis of the hip (TOH), are thought to be the result of the failure of the normal metabolic adaptations of pregnancy albeit without any definitive understanding of the underlying pathology [6,7].

Although rare, PIO can affect not only the development of associated vertebral fractures but also potentially alter sagittal

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## Author's Photo Gallery



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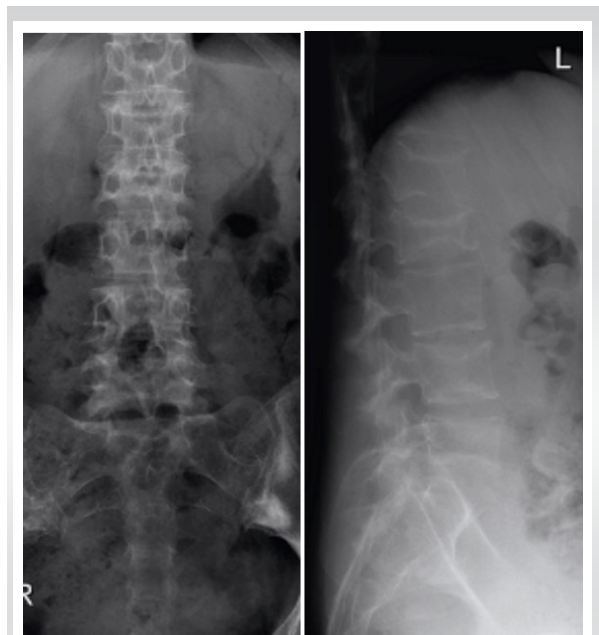
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**Figure 1:** Plain film radiographs in anterior-posterior (left) and lateral (right) views at initial presentation to the emergency department.



**Figure 2:** Computed tomography scan of lumbar spine in sagittal view performed under spinal specialist.

patient, she was keen to progress with conservative management of her numerous fractures. No risk factor or predisposing condition was identified, and the patient's dietary assessment reflected adequate calcium intake. Serological laboratory investigations demonstrated low 25 (OH) Vitamin D levels (26 nmol: Normal range >50). However, the interval plain film radiographs performed at the spinal clinic 2 weeks later showed the progression of her lumbar spine into kyphosis and positive sagittal balance.

Thereafter, a computed tomography scan revealed biconcave compression fractures of the T9, T11, T12 (all with 20% loss of height), L1 (30%), L2 (50%), L4 (30%), and L5 (40%) vertebrae

imbalance, both of which can have extremely disabling consequences in young female patients [8]. Furthermore, the rarity of such presentations means that optimal management of such fractures presents a unique challenge to the attending surgeon, with treatment focused on potential prevention and/or correction of resulting deformities. Therefore, the primary aim of this study sought to report our findings in a young patient who presented with PIO.

### Case Report

A healthy nulliparous (G1+0) 27-year-old female with no previous medical history presented to the emergency department early in her second trimester after developing severe acute onset lower back pain, in the absence of any history of trauma. On clinical examination, it was reported that she displayed point tenderness at the level of the thoracolumbar junction, and difficulty ambulating secondary to pain, with no evidence of any radiculopathy or lower limb neurological deficits. Plain film radiographs were performed in anteroposterior and lateral views of her lumbar spine alone demonstrating compression fractures of her 1st (30%) and 2nd (50%) lumbar vertebrae (Fig. 1).

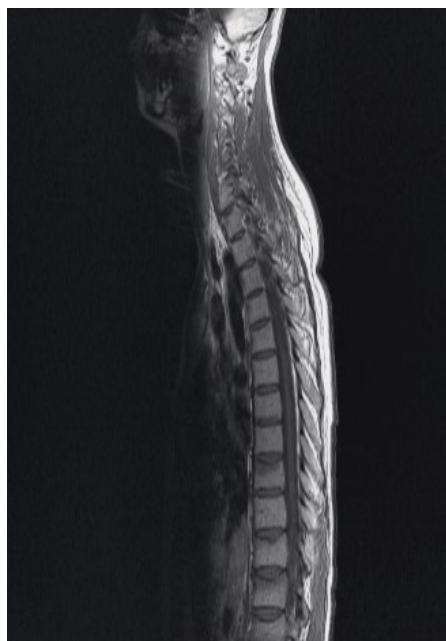
She was subsequently referred to the fracture clinic at 1-week postpartum, where she was initially treated in a soft thoracolumbar spinal orthosis (TLSO) with an associated referral to our institution's board-certified, fellowship-trained orthopedic spinal surgeon at 3-week postpartum. Although the various management options were discussed at length with our

(Fig. 2).

The middle and posterior columns remained intact at each vertebral level [9]. There was no evidence of any involvement of the posterior elements on magnetic resonance imaging and short-TI Inversion recovery sequence confirmed the fractures to be recent. She had a low pelvic incidence, low lumbar lordosis, and a compensatory hypokyphosis of the thoracic spine (Fig. 3 and 4). A summary of the sagittal balance parameters is shown in Table 1.

A dual-energy X-ray absorptiometry (DEXA) scan was subsequently performed diagnosing gross osteoporosis in the lumbar spine with T-scores ranging from -4 to -4.4 (converted to Z-scores of -3.9 to -4.3), notably more severe than measurements at the femoral head. The results of the DEXA scan are further illustrated in Table 2.

Treatment with bracing was continued and in consultation with the bone health service (which in our institution includes specialist endocrinology and rheumatology services), further actions implemented with the aim of halting bone loss included cessation of breastfeeding. In an effort to combat potential exacerbation of her now-known severe osteoporotic condition, the patient was thereafter commenced on a 24 month course of 20 mg of teriparatide once daily subcutaneously and the addition of calcium carbonate 500 mg and Vitamin D3 800 international units combined once daily orally. All potential benefits and risks of such treatment were outlined in full before commencement.



**Figure 3:** Computed tomography scan of cervical and thoracic spine in sagittal view performed under spinal specialist.



**Figure 4:** Magnetic resonance imaging performed under a spinal specialist.



**Figure 5:** Plain film radiographs in scoliosis erect lateral view at interval follow-up with a spinal specialist.

At 4-month follow-up, she reported improvement in back pain with no myelopathic or radicular features, with no neurological deficits reported globally on examination. In addition, her TLSO brace was weaned to long-distance mobilization only, with a subsequent referral for physiotherapy to strengthen core stability exercises as well as tolerated. Prominent kyphosis at the thoracolumbar junction remained evident on clinical examination and imaging. The low lumbar lordosis and compensatory low thoracic kyphosis both remained stable through the follow-up period.

Further improvement in back pain and increased mobility were reported at 7-month follow-up and repeat imaging showed that lumbar lordosis, thoracic kyphosis, and sagittal parameters were unchanged from the previous. Serological laboratory levels of 25 (OH) Vitamin D had normalized (76 nmol/L) at this point. No surgical intervention was required throughout the patient's clinical course. The patient was successfully discharged from the service at 18-month follow-up following counseling regarding potential long-term sequelae and the option for review as needed, with a prescription of 6 monthly injections of denosumab. At 6-year follow-up (Fig. 5), her latest DEXA scan reported moderate osteopenia, with a T-Score of  $-1.5$ . At the final follow-up, the patient was asked for permission that data related to this case would be converted into anonymized form with the intention of publication. The patient subsequently provided informed consent in written form for this use.

## Discussion

### Epidemiology

While the incidence of PIO is debated in the literature, however, consensus exists that spinal involvement represents a rare sequelae, with a reported incidence of approximately 0.4/100,000 [10, 11]. PIO classically presents in primigravid women in the first 3-month postpartum [12], with acute onset back pain, but up to 40% of those affected will at the time or retrospectively describe symptoms during the third trimester [13]. Furthermore, neither lactation nor multiple pregnancies have been shown to be associated with subsequent osteoporosis [14], nor is there a reported association between the duration of lactation and fracture risk [15]. However, studies demonstrate that a proportion of patients with PIO may have a genetic predisposition to low peak bone mass, which may contribute to their increased risk of fracture during pregnancy [16], while the presence of premenopausal fractures has been shown to increase the patient's risk of postmenopausal fracture by 35% [17]. Furthermore, a combination of congenital and acquired risk factors has been identified for PIO, namely, the presence of low body weight, family history, Vitamin D deficiency, and smoking [18]. A retrospective study performed over a 20-year period by O'Sullivan et al. [19] found that in their 11 patient series, over 80% of patients had at least one risk factor for osteoporosis, despite their young age and pre-menopausal

Sagittal parameters at presentation	Value
Sagittal vertical axis	5.8 cm
Pelvic incidence	31°
Pelvic tilt	15°
Sacral slope	16°
Lumbar lordosis (L1–S1)	14°
Lumbar lordosis (L4–S1)	28°
Lumbar lordosis (L1–L3)	20°
Thoracic kyphosis	10°
T9 PA	9°

cm: Centimeters, L: Lumbar, PA: Posterior-Anterior

**Table 1: Sagittal parameters at presentation.**

status.

### TOH

Although uncommon, TOH represents a more common clinical phenomenon to PIO which is noted to be more prevalent in the male population [20]. Many known risk factors for TOH match those pre-disposing individuals to osteoporosis, including low serum Vitamin-D levels, steroid usage, and low circulating phosphate and calcium levels [21, 22]. First reported by Curtiss and Kincaid in 1959, pregnancy was noted to represent a risk factor for TOH in cases whereby the young female patient is affected, with significant demineralization of four proximal femurs [23]. With respect to both PIO and TOH, previous literature has found that although the pregnant lady is at increased risk of reduced bone density, any associated bone loss is believed to be physiologically reversible in the postpartum period [24]. Furthermore, previous series have found satisfactory clinical outcomes for both osteoporotic vertebral compression fractures (OVCFs) and TOH, with little postpartum morbidity reported in the long term [19,20].

### Sagittal imbalance

The sagittal alignment, or “front to back balance,” of a normal spine is directly related to physiological spinal alignment by the muscular forces [25]. Therefore, sagittal balance is dependent on the positional relationship between the thoracic curve, lumbar curve, sacrum, and the pelvis. A normal sagittal vertical

Bone	Z-Score	T-Score	BMD
L3	-4.3	-4.4	0.673 g/cm <sup>2</sup>
L4	-3.5	-3.6	0.774 g/cm <sup>1</sup>
L3-L4	-3.9	-4.0	0.726 g/cm <sup>0</sup>
Right Femur Neck	-1.9	-2.0	0.747 g/cm <sup>1</sup>
Left Femur Neck	-1.9	-2.0	0.747 g/cm <sup>2</sup>

BMD: Bone mineral density, DEXA: Dualenergy X-ray absorptiometry, g/cm<sup>2</sup>: grams/centimeter, L: Lumbar

**Table 2: Initial DEXA Scan Result.**

axis is reflected in the plumb line lying within 3 cm of the sacral promontory (anteriorly or posteriorly); however, this balance may be altered positively (plumb line lies >3 cm anteriorly) or negatively (plumb line lies >3 cm posteriorly [26]. Such imbalances may be due to intrinsic alignment issues or changes in alignment due to any number of musculoskeletal pathologies, such as PIO in the case of the lady previously discussed in this case.

### Management

The management of osteoporosis in the general population is well established, with specific involvement of appropriate investigations for any underlying secondary cause, including Vitamin D deficiency, coeliac disease, anorexia nervosa, mastocytosis, and hyperparathyroidism and hyperthyroidism [27]. However, as a paucity of prospective literature exists for cases of PIO, optimal treatment strategies for such cases remain controversial [28]. Recommendations for initial avoidance of strenuous activities, appropriate analgesia and physiotherapy as well as appropriate re-mineralization with calcium and Vitamin D supplementation tend to be incorporated into medical management in the majority of cases [29]. Although discrepancies in opinion exist among obstetricians and pediatricians globally, logical cessation of lactation in cases of PIO reduces the deleterious effect on the bodies’ calcium stores, with these initial steps alone have been shown to result in complete resolution of symptoms at 4 months [30].

Regarding further pharmacological treatment, there is a far weaker consensus. In all cases, a major concern that exists when treating gravid women evolves around potential teratogenicity or implications fetal development [31, 32]. Both bisphosphonates and teriparatide fall into the United States Federal Drug Administration’s Category C, theoretically rendering them contra-indicated during pregnancy unless there is a strong clinical argument for their use [33]. Stumpf et al. previously demonstrated that in the absence of an underlying



cause, bone mineral density will return toward previous levels post-pregnancy granted that suitable osteoanabolic therapy is utilized [34]. In addition, consideration as to whether bisphosphonates or teriparatide should be used in these cases is debated; teriparatide has previously been reported that result in a greater positive effect on bone mineral density when compared to bisphosphonates in PIO [35], with lower reported pain scores than those treated with percutaneous kyphoplasty at short-term follow-up [36].

### Non-operative orthopedic management

The use of bracing as an acute non-operative treatment for OVCFs is well established, but it remains largely opinion based, with a lack of underpinning evidence which is majorly based on their application in the management of acute, traumatic burst fractures [37]. In their meta-analysis of four randomized control trials, Jin and Lee suggested that bracing plays a role in significantly improving patient-reported pain scores and quality of life when compared to controls in patients with OVCFs [38], although two of the included trials were not supportive of bracing in such patients [39,40]. Theoretically, bracing decreases the load over the anterior spinal column by attempting to reduce forward flexion, therefore decreasing the painful contraction of erector spinae which occurs with the sagittal imbalance of multiple vertebral compression fractures [41]. Typically prescribed for a 3-month period, the use of bracing has previously been reported to prevent new or progression of old fractures at 12-month follow-up in young osteoporotic female patients suffering PIO, particularly in cases of thoracolumbar compression fractures when combined with supplementary medical and physical therapy [8,42-44]. Our patient was treated in a soft TLSO brace, although acknowledgment of the study Kim et al. [40] is warranted, who demonstrated that there were no significant differences in reported pain scores at 3-month follow-up between patients managed with a soft brace, a hard brace, or no brace at all.

Although rare, our patient's presentation with seven vertebral fractures is not unique. A previous study by Ofluoglu and Ofluoglu [45] reported the conservative management of a 30-year-old patient with PIO presenting with eight OVCFs in her spine, with acceptable results at 12-month follow-up. For their

patient, the option of balloon kyphoplasty was considered with the patient; however, the concern of potential difficulty performing an osteotomy in future through a cement-filled vertebral body tipped the balance away from operative orthopedic management in her case. Osteotomy was held in reserve for our patient until well after the peripartum period in case potential symptomology or sagittal malalignment became unsatisfactory in our patient's case in future. This option provided a potentially less demanding approach should operative intervention have been required, in comparison to a potential hypothetical situation in which a failed trial of kyphoplasty has altered native anatomy. Furthermore, our patient has reported satisfactory outcomes at 6-year follow-up, with occasional mild lumbar back pain on exertion. Having been successfully discharged from our service, she is continuously being administered therapeutic denosumab on a bi-annual basis which is followed up in the community. Her latest DEXA scan reported a moderate osteopenia, with a T-Score of  $-1.5$  (Z-score of  $-1.4$ ).

### Conclusion

Although rare in incidence, PIO-associated OVCFs represent a significant cause of morbidity for the young, peripartum female population. The lady in our case is an example of how multiple PIO-associated OVCFs may be managed successfully with a combination of medical and non-operative orthopedic therapies with medium-term follow-up. However, one must always consider the significance of sagittal imbalance and the potential role of operative orthopedic intervention or even correction, particularly when managing such a young patient cohort with such a severely morbid disease process such as osteoporosis.

### Clinical Message

The lady in our case is an example of how multiple PIO-associated OVCFs may be managed successfully with a combination of medical and non-operative orthopedic therapies with medium-term follow-up; however, the significance of sagittal imbalance must be a consideration when electing either management option.

**Declaration of patient consent:** The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

**Conflict of interest:** Nil    **Source of support:** None

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