

An Atraumatic Sacral Fracture with Lumbosacral Radiculopathy Complicating the Early Postpartum Period: A Case Report

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
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Conflict of interest: None declared

Patient: Female, 26
Final Diagnosis: Atraumatic fatigue sacral fracture
Symptoms: Lumbar back pain with radiculopathy
Medication: —
Clinical Procedure: —
Specialty: General and Internal Medicine

Objective: Rare disease

Background: Sacral stress fractures are rare complications of pregnancy and the early postpartum. Of these, few present with lumbosacral radiculopathy. We report the first Australian case of a young multiparous woman who sustained an atraumatic, fatigue sacral fracture with associated radiculopathy. We highlight the diagnostic process and chronic management of this case, particularly in relation to a future pregnancy.





Case Report: A 26-year-old multiparous Caucasian female presented with worsening lumbosacral back pain and radicular symptoms following the rapid and spontaneous vaginal delivery of her second infant. Her pregnancy was unremarkable and she had no personal risk factors for osteoporosis. A magnetic resonance imaging (MRI) scan confirmed the diagnosis of a right S1 vertebral fracture. Bone densitometry and fasting bone metabolic testing excluded pregnancy-associated osteoporosis. She was managed conservatively with intermittent bed rest, regular physiotherapy and multimodal analgesia. During a future pregnancy, she experienced a severe exacerbation of her lumbosacral radiculopathy requiring hospital admission, up-titration of her analgesia and a right S1 epidural injection. She subsequently underwent an elective caesarean section and has since benefitted from regular hydrotherapy.

Conclusions: Lumbosacral radiculopathy in the absence of trauma during pregnancy or the early postpartum should prompt consideration of an underlying atraumatic, fatigue sacral fracture. Such fractures may result from the abnormal biomechanical loading of the sacrum during rapid vaginal deliveries and are most effectively diagnosed by MRI. Conservative management strategies involving physiotherapy and multimodal analgesia are recommended. Future pregnancies may exacerbate radicular symptoms. Such patients may subsequently benefit from elective caesarean section deliveries and hydrotherapy.

MeSH Keywords: Postpartum Period • Radiculopathy • Spinal Fractures

Abbreviations: **bid** – bis in die; **cm** – centimeter; **CT** – computerized tomography; **G** – gravida; **g** – grams; **kg** – kilogram; **L** – liter; **MRI** – magnetic resonance imaging; **m** – meter; **mL** – milliliter; **nmol** – nanomoles; **P** – para; **prn** – pro re nata; **%** – percent

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Background

Calcium requirements during pregnancy and the immediate postpartum represent a period of maternal metabolic stress. The developing fetus demands high levels of calcium which is predominantly obtained through intestinal absorption of maternal calcium during the first and second trimesters [1,2]. During the third trimester, fetal requirements often exceed maternal supplies of calcium obtained from the intestine. Subsequently, increased bone resorption mediated through 1,25-hydroxyvitamin D and osteoclastic activity is facilitated to meet this fetal demand [1]. Studies have shown that the third trimester is dominated by increased serum markers of bone resorption (i.e., pyridinoline, deoxypridinoline and N-telopeptide) and reduced bone mineral density of the femoral neck, lumbar spine, and forearm [1]. Upregulation of osteoblastic biomarkers is also noted before birth (i.e., bone specific alkaline phosphatase, procollagen extension peptide) [1]. Consequently, extensive maternal bone remodeling occurs during the third trimester to support ongoing fetal growth and calcium requirements.

Although uncommon, pregnancy-associated osteoporosis may occur as a consequence of deranged calcium homeostasis and potentially predispose the pregnant mother to an increased risk of fractures [2]. Fractures of the spine and pelvis following minimal trauma in pregnant mothers with osteoporosis have been previously described [3,4]. However, atraumatic stress fractures of the sacrum arising intrapartum or during the early postpartum are unusual and rare complications of pregnancy. To date, only a handful of cases having been reported across North America [5–7], Europe [8–13], the Middle East [14–19], and South Korea [20]. These fractures are classified as either insufficiency or fatigue fractures [21]. Insufficiency fractures, which are most commonly linked with pregnancy-associated osteoporosis, occur when minimal energy disrupts a bony matrix lacking appropriate mineralization and elastic resistance. In contrast, fatigue fractures occur in the non-osteoporotic setting and result from repeated forces applied to normal bone that eventually exceeds its elastic threshold. The majority of reported cases are representative of fatigue sacral fractures sustained during the early postpartum in multiparous women, irrespective of the method of delivery [5,8,15–17,19].

We report the first Australian case of a 26-year-old multiparous woman sustaining an atraumatic, fatigue sacral fracture with associated lumbosacral radiculopathy in the early postpartum. Further, we are the first to demonstrate the chronic, relapsing effect that a future pregnancy had on this patient's radicular symptoms, management thereof and overall morbidity. The case highlights the importance of sacral pain with radiculopathy as key red flag symptoms that should prompt the consideration of an underlying sacral fracture in the early postpartum.

Case Report

A 26-year-old Caucasian, multiparous (G₃P₂) female, previously fit and well, presented 4 days following the spontaneous vaginal delivery of her second child with worsening lower back pain and immobility. The patient described a deep, dull ache involving the right sacral paraspinal region. Moreover, she reported radiation of a sharp pain down the posterior component of her right thigh to her heel. She complained of worsening paresthesia affecting her right foot and toes. The pain was exacerbated by sitting, prolonged weight bearing and lifting of objects. She denied any bowel or bladder incontinence. She reported that she had experienced lumbosacral pain since the 25th week of her pregnancy, but this had acutely worsened since the birth of her second child. On examination, tenderness was elicited with palpation over the right paraspinal sacrum with associated voluntary guarding. The patient maintained her right hip in fixed flexion at 20 degrees and complained of pain with extension. The power in her right hip and leg were grossly reduced secondary to pain. Light touch sensation involving the dorsum and plantar surfaces of her right foot were diminished. A straight leg raise test was positive at 20 degrees.

The patient's pregnancy was essentially unremarkable with only evidence of a bicornuate uterus being noted during ultrasonography. Routine antenatal blood and urine testing were unremarkable, with an adequate vitamin D level of 75 nmol/L. The patient had a previous history of migraines, an appendectomy and a previous missed miscarriage for which she had an uncomplicated dilatation and curettage 4 years earlier. Her family history was significant for maternal osteoporosis and type 2 diabetes mellitus. She only took iron supplements daily and did not smoke or drink alcohol. She undertook regular, low intensity walking exercises up to 2 months prior to her delivery but did not partake in any strenuous sports or running. She had a balanced diet with an adequate intake of dairy products and was not vegetarian. She had not received heparin previously. During her first pregnancy, she only breastfed for 8 weeks and her infant weighed 2700 g at birth.

Concern was noted for potential intrauterine growth restriction (10th percentile) during this pregnancy and the patient was induced with a Foley's catheter at 38⁺⁵/40 weeks gestation. Her anthropometric measures noted a weight of 76 kg (gain of 9 kg during pregnancy), height of 173.5 cm and body mass index (BMI) of 25.2 kg/m². Her labor was augmented with Syntocin and she received an uncomplicated epidural comprising of a 15 mL volume of 0.125% buprenorphine and 5 mg/mL fentanyl into the L3/4 space. She was noted to have some hypotension post insertion of the epidural, which was managed with 1.0 mg of intravenous metaraminol. The infant had a cephalic presentation and left occiput anterior position. The labor proceeded rapidly, with the first and second stages

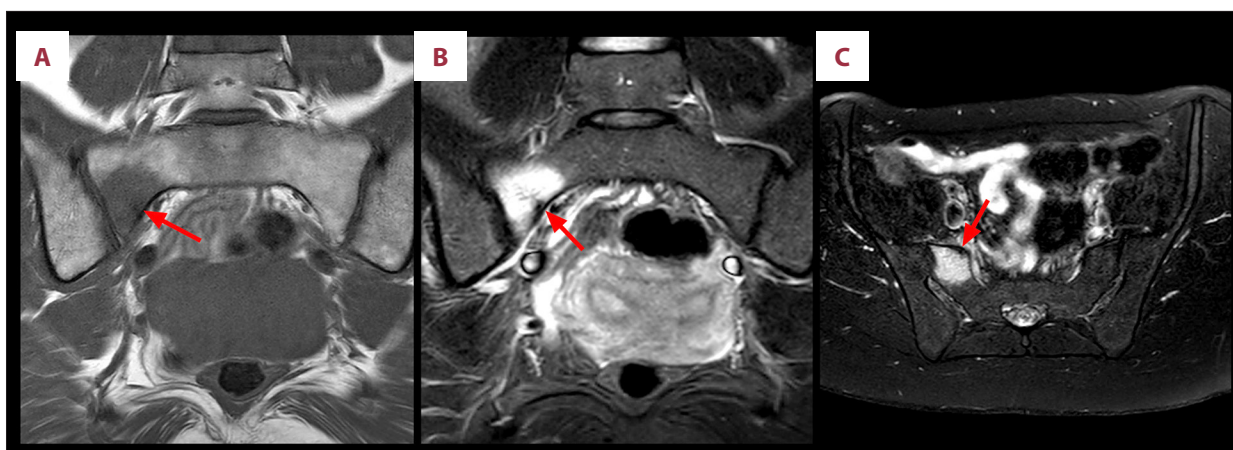


Figure 1. Magnetic resonance imaging (MRI) of the patient's lumbosacral spine at diagnosis of the sacral fracture. (A) T1-weighted, (B) STIR and (C) T2 SPAIR imaging showing focal marrow edema anteriorly (arrows) in the sacrum at the level of S1. Features are consistent with a stress fracture of the right S1 segment.

lasting 180 minutes and 32 minutes respectively. A male infant was born by spontaneous vaginal delivery not requiring intervention with forceps. He did not have shoulder dystocia and the patient had no perineal lacerations. The infant's anthropometric measures noted a birth weight of 3045 g, length of 51.5 cm, and head circumference of 35.0 cm. The patient did not breastfeed this infant and was discharged.

Upon readmission to hospital for her worsening lower back pain and associated sciatica, regular and multimodal analgesia with physiotherapy were implemented with minimal benefit. The patient failed to improve and anesthetic review was obtained. Differential diagnoses for lumbosacral pain with associated radiculopathy were considered, including an epidural related nerve injury, lumbosacral plexopathy, sacroiliitis, or other pathologies (e.g., discogenic pathology, spinal stenosis, fractures). A magnetic resonance imaging (MRI) scan of the lumbosacral spine was performed 2 weeks post-partum which noted focal marrow edema involving the right S1 level and an equivocal anterior cortical fracture of the S1 vertebra (confirmed with a repeat MRI scan 3 weeks later, Figure 1). A computed tomography (CT) scan undertaken at the same time could not appreciate the fracture.

Multidisciplinary reviews were undertaken involving an orthopedic surgeon, anesthetist, and general physician over a 1-month period following the patient's second delivery. Further examinations revealed no evidence of chronic orthopedic or rheumatological disease. Clinical features of vasculitis, thyroid disease, and other endocrinopathies were also absent. Comprehensive blood testing (Table 1) and bone mineral densitometry were unremarkable, with Z-scores of the hip and spine reported at +0.5 and +0.6 respectively. The patient was subsequently diagnosed with an atraumatic fatigue fracture of the sacrum with associated radiculopathy. Conservative management was

recommended and comprised of intermittent best rest, regular physiotherapy, and weight bearing as tolerated. A multimodal analgesic regimen involving regular pregabalin and paracetamol, with pro re nata (PRN) celecoxib and immediate-release tapentadol was prescribed. The patient reported the greatest pain-relieving effect from pregabalin. Over the next 5 months, her pain resolved and pregabalin was ceased. Occasionally, she reported bouts of exacerbated back pain and radiculopathy with minimal exercise, but this settled with rest and PRN anti-neuropathic analgesia.

Two years later, she fell pregnant with her third child. At approximately 34 weeks gestation, she was admitted to hospital following a severe exacerbation of her sacral radicular pain that had compromised her mobility and ability to transfer out of bed. She was acutely managed with dexamethasone 2 mg bid for 5 days in addition to temporarily increasing her pregabalin dose. She later had an ultrasound-guided right S1 epidural injection to settle her pain. Repeat MRI of her lumbosacral spine noted no new fractures and resolution of her previous S1 fracture (Figure 2). She later underwent an elective lower uterine Caesarean section without complication, giving birth to a healthy infant weighing 2825 g. Ongoing hydrotherapy was commenced post-delivery with positive effect and presently, her lumbosacral radiculopathy is well managed with low dose pregabalin and regular physiotherapy intervention.

Discussion

Lower back pain related to pregnancy is common and affects >50% of women [16]. The vast majority result from hyperlordosis of the maternal spine as the fetus grows. In the postpartum, lumbosacral pain may persist and is usually related to ligamentous strains of the pelvis sustained from the delivery

Table 1. Extensive hematological, urine and biochemical results at the time of diagnosis of the patient's sacral fracture.

Parameter	Units	Reference	Result
Fasting metabolic bone study			
Serum calcium (ionized at pH 7.40)	mmol/L	1.12–1.30	1.17
Serum calcium (total)	mmol/L	2.20–2.55	2.26
Serum albumin	g/L	38–50	41
Serum calcium (corrected)	mmol/L	2.20–2.55	2.28
Serum magnesium	mmol/L	0.70–1.20	0.88
Serum phosphate	mmol/L	0.80–1.50	1.40
Serum creatinine	µmol/L	30–100	72
Estimated glomerular filtration rate (EGFR)	mL/min/1.73 m ²	>60	>90
Serum gamma glutamyltransferase (GGT)	U/L	<31	5
Plasma intact parathyroid hormone	pmol/L	1.50–8.00	5.20
Serum 1,25-hydroxyvitamin D	nmol/L	>50	83
Urine calcium/creatinine ratio	mmol/mol	100–580	194
Urine calcium excretion	µmol/L*	9–42	14
Renal phosphate threshold	mmol/L*	0.75–1.35	1.35
Serum alkaline phosphatase (ALP)	U/L	20–105	67
Urine N-telopeptide/creatinine (NTX)	BCE/mmol	<50	33
Full blood profile			
Hemoglobin	g/L	115–155	139
White cell count	×10 ⁹ /L	4.0–11.0	4.6
Platelets	×10 ⁹ /L	150–400	210
Miscellaneous results			
Ferritin	µg/L	30–200	119
Transferrin saturation	%	15–55	16
Thyroid stimulating hormone (TSH)	mU/L	0.40–4.00	0.86
Vitamin B12	pmol/L	150–750	313
Prolactin	U/L	<500	230

* Urine calcium excretion calculated as a function of the glomerular filtration rate.

of the infant [8]. However, fractures of the sacrum intrapartum or shortly after birth are rare complications of pregnancy and cause progressive immobility. The underlying pathogenesis is variable, with insufficiency fractures being related to pregnancy associated osteoporosis [7,18,20]. However, most atraumatic sacral fractures are representative of fatigue fractures and occur in women without osteoporosis [5,6,8–17,19]. It is postulated that repeated forces applied to the maternal sacrum eventually exceed its elastic threshold and cause stress

fractures [21]. Lumbosacral radiculopathy is rarely associated with these fractures and its overall incidence is estimated at 0.08% [16]. To the best of our knowledge, we have reported the first Australian case of a young multiparous woman with an atraumatic, fatigue sacral fracture with associated lumbosacral radiculopathy.

Several risk factors that predispose women to atraumatic sacral fractures in the intrapartum and early postpartum have been



Figure 2. Magnetic resonance imaging (MRI) of the lumbosacral spine two years following the initial diagnosis of the patient's sacral fracture. (A, B) T1-weighted and (C) T2 SPAIR imaging showing resolution of the previous right S1 stress fracture (arrows). Mild sclerosis at the site of the previous is appreciated on the axial T1-weighted image (B).

identified. These include excess weight gain and strenuous exercise during pregnancy, severe hyperlordosis, osteoporosis, lactation, macrosomia, rapid spontaneous vaginal deliveries, forceps deliveries and anticoagulation with heparin [5–20]. The only risk factor applicable to our patient was that she had a rapid spontaneous vaginal delivery. She did not gain excess amounts of weight, delivered a normal baby and did not undertake strenuous exercise. Of interest, only Sibila et al. [8] have published a similar case in a 31-year-old multiparous woman with severe lumbar back pain and immobility shortly after she vaginally delivered a small infant within 20 minutes. It is hypothesized that the rapidity of spontaneous vaginal deliveries exerts undue stress on the sacrum, thereby altering its biomechanical loading properties and inducing stress fractures [8,16]. In parallel with other cases [5,8,11,15,16,19], our patient reported the onset of severe sacral pain within the first two weeks following her labor. However, only 3 other cases [8,16,20] reported radicular symptoms, with 1 patient being misdiagnosed with discogenic disease. Although lower back pain in the early postpartum is common, especially after spontaneous vaginal deliveries, a clinical course of worsening pain, immobility and associated radicular symptomatology should necessitate further investigation for an underlying sacral fracture.

Upon clinical suspicion of an underlying sacral fracture, the diagnosis should be confirmed by MRI. Often, fatigue fractures will show surrounding marrow edema with formal radiographic evidence of the fracture only becoming apparent within 1 month of the onset of symptoms (Figure 1). We and others have shown that CT and x-rays lack the appropriate resolution to isolate such fractures and pose teratogenic risks to the fetus [7,11–13,16,17,20]. Exclusion of pregnancy associated osteoporosis is also required by means of formal bone densitometry and fasting metabolic biochemical assessments [7,15–17,20]. Although fatigue fractures of the sacrum arise in the non-osteoporotic setting, previous case reports have noted reduced bone density in affected women

(e.g., Z-score -1.60 [19]). It is postulated that transient osteoporosis of pregnancy and the early postpartum increases the risk of fractures and occurs due to lactation, hyperprolactinemia and increased relaxin levels weakening the pelvic ligaments [11,16]. The fracture risk is further augmented by deranged maternal calcium homeostasis and increased bone remodeling during the third trimester and immediate postpartum [1,2]. One may argue that “fatigue” sacral fractures with densitometry results illustrating features of osteopenia without osteoporosis (e.g., Z-scores: -0.5 to -1.9) should be reclassified as a combined “fatigue-insufficiency” fracture. However, and in contrast with all other reports [5–20], our patient demonstrated above normal bone density findings. This suggests that our patient sustained a purely fatigue related fracture of her sacrum secondary to altered biomechanical loading forces applied during her rapid spontaneous vaginal delivery.

Intermittent bed rest, physiotherapy, and multimodal analgesia form the foundations of conservatively treating atraumatic sacral fractures in the postpartum. Importantly, early weight bearing should be encouraged as external bone loading forces stimulates bone growth and recovery [18]. Öztürk et al. [18] have also shown that transcutaneous electrical nerve stimulation and pulsed electromagnetic field therapy may offer analgesic properties and facilitate bone healing. Our patient and others have reported resolution of their symptoms within 6 months of conservative treatment [5–20]. However, the morbidity of sacral fractures in future pregnancies and their subsequent management has not been previously considered. We have demonstrated how a future pregnancy exacerbated our patient's radicular symptoms, especially in the third trimester. The up-titration of anti-neuropathic agents, implementation of steroid therapy and consideration of targeted epidural analgesia may provide positive clinical benefits to such patients. To avoid postpartum exacerbations of established sacral fracture radiculopathy, we recommended our patient underwent an elective caesarean section with post-delivery hydrotherapy.

Previous studies have shown improved analgesic and quality of life outcomes conferred by hydrotherapy in patients with osteoarthritis and osteoporosis [22,23]. Therefore, it is worthwhile considering hydrotherapy in the conservative and chronic management paradigm of atraumatic sacral fractures with associated radiculopathy.

Conclusions

Atraumatic sacral fractures arising intrapartum or during the early postpartum are rare complications of pregnancy and represent a heterogeneous clinical course. Lumbosacral radiculopathy is a rare feature of these fractures and represent a key red flag symptom. MRI represents the gold standard modality to diagnose such fractures, but formal bone densitometry and fasting bone metabolic examinations must be performed to exclude osteoporosis. We recommend treating postpartum patients with atraumatic sacral fractures with multimodal analgesia, physiotherapy, intermittent bed rest, and early weight bearing.

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Exacerbations of lumbosacral radiculopathy may occur with future pregnancies and can be appropriately managed with elective caesarean deliveries. Hydrotherapy may further assist recovery and the chronic maintenance of radicular symptoms.

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Conflicts of Interest

None.