

Case Report: Transient Osteoporosis of Pregnancy in the Knee

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Abstract

Transient osteoporosis of pregnancy is an uncommon skeletal pathology that develops during the third trimester in otherwise healthy pregnancies. When it rarely presents, it is often cited in the literature with primary pathology in the hip of pregnant patients. We present the diagnostic evaluation and management of a case of transient osteoporosis developing in the knee in the last months of pregnancy, which is an unusual location for an already rare skeletal pathology.

Keywords: *Transient Osteoporosis of Pregnancy (TOP); Pregnancy; Knee*

Introduction/Background

Transient osteoporosis of pregnancy (TOP) is a rare condition first described in the medical literature in 1959 in a report of three cases of pain in the hip or thigh that developed in the third trimester of pregnancy with confirmation of demineralization in the femoral heads on x-ray [1]. Despite the rarity of TOP, its relevance is significant to obstetricians and orthopedists alike as it affects healthy females in otherwise uncomplicated pregnancies. The condition itself is secondary to temporary bone loss with a self-limited disease course that resolves postpartum.

Clinical presentation involves complaints of sudden pain without recent trauma. Given the rarity of the disease, in addition to the non-traumatic patient presentation, diagnosis can be difficult in the early stages secondary to physician lack of awareness or clinical suspicion. Diagnostic imaging often confirms this diagnosis of exclusion [2,3]. Ultimately, management of transient osteoporosis is with conservative measures until delivery and resolution of the disease often occurs spontaneously in the postpartum state [2,3]. Identifying this case report will lead to earlier diagnoses, ultimately reducing unnecessary healthcare costs with imaging and treatments for mimicking diseases. Having an early and accurate diagnosis will allow patients to understand their disease progression and allow physicians to manage their antenatal course appropriately.

Case Presentation

A 36 year old Caucasian woman, gravida 3, para 2002, at 31 weeks and 0 days gestational age with dichorionic diamniotic twin intrauterine pregnancy presented to the emergency department for right knee pain that started a few days prior to her arrival, worsening

with weight bearing. She denied any recent falls, injuries, or trauma. The patient denied a history of drinking alcohol or smoking tobacco products, although she admitted to marijuana use in pregnancy. She was not taking prescribed anticonvulsants or corticosteroids. She did not have a history of any metabolic or malabsorption disease. Her surgical history is significant for prior partial thyroid lobectomy and two prior Cesarean sections. Vitals in the emergency room were notable for tachycardia in the 100s but blood pressure was normotensive and she was afebrile with a normal respiratory rate. Patient’s BMI was 32. On physical exam, her right knee had no gross deformities, joint effusion, overlying erythema or warmth however it was noted to be tender to palpation at the medial aspect. The patient was offered an x-ray at the time but she declined because there was no traumatic injury. She was subsequently placed in a knee immobilizer and discharged home for outpatient follow up.

Ten days later, she presented to the obstetric emergency department for evaluation due to significantly worsening pain and swelling in her right leg. Evaluation was negative on lower extremity dopplers and she was discharged to home with initiation of enoxaparin for venous thromboembolism prophylaxis. It was recommended that she follow up with outpatient orthopedics for evaluation. The patient was unable to follow up outpatient with the referred orthopedist due to her insurance coverage.

The patient then presented to the obstetric emergency department at 33 weeks and 2 days gestational age with a complaint of intermittent lower pelvic pain in addition to persistent knee pain, unchanged from prior evaluations. She was found to be contracting regularly on tocometry and subsequently admitted to the hospital for 23 hour observation with concern for preterm labor. As the patient continued to complain of severe right knee pain and because she was unable to complete outpatient evaluation with orthopedics, an inpatient consultation was placed. X-rays of the right knee were significant for diffuse osteopenia in the distal femur and proximal tibia (Figure 1 and 2). Follow up MRI demonstrated intense bone marrow edema throughout the medial femoral condyle and to a lesser extent along the lateral femoral condyle. Additionally, a subtle T1 hypointense line was identified along the medial femoral condyle (Figure 3). Findings were suggestive of transient osteoporosis of the knee with superimposed stress fracture.



Figure 1: Anterior posterior view of right knee x-ray.



Figure 2: Lateral view of the right knee x-ray.



Figure 3: MRI of right knee demonstrating intense bone marrow edema noted throughout the medial femoral condyle and to a lesser extent along the lateral femoral condyle. A subtle T1 hypo intense line is noted along the medial femoral condyle, likely representing a stress fracture. Findings could be the sequela of transient osteoporosis of the knee with a superimposed stress fracture.

The patient was ultimately discharged with improvement of her preterm contractions with strict instructions to remain non-weight bearing for the remainder of her pregnancy. The plan was for scheduled repeat low transverse Cesarean section at 38 weeks, however the patient presented to the triage unit at 35 weeks and 3 days with multiple complaints. Ultimately, fetal heart tracing demonstrated a per-

sistent category II tracing for fetus B with recurrent decelerations and the decision was made to proceed with a Cesarean delivery. Twin A was delivered with Apgars of 8 and 9 at 1 and 5 minutes respectively. Twin B was delivered with Apgars of 7 and 8 respectively. Initial evaluation of both twins by neonatology revealed normal musculoskeletal growth with appropriate range of motion of all extremities.

The surgery was uncomplicated and the patient met all her postoperative milestones postpartum. While hospitalized, she was evaluated by occupation and physical therapy services. She was instructed to follow up outpatient with both obstetrics and orthopedics, but unfortunately she did not attend her 1 week postpartum incision check or routine 6 week postpartum visit.

Discussion

As in our case presentation, TOP typically presents suddenly with unilateral onset of pain without an inciting trauma in the third trimester of pregnancy [2]. While exact pathophysiology for TOP is unclear, the majority of the literature suggests that during pregnancy, if maternal intake of calcium is insufficient, the maternal skeleton will undergo resorption to supplement the needs of the growing fetus [4]. A single case control study in the literature by Hadji, *et al.* suggests TOP is a multifactorial disease with associations secondary to immobility, dental problems, and lack of exercise in childhood [3]. A literature review demonstrates a mean maternal age of onset of TOP to be 32.1 weeks of gestational age [5]. Of these reviewed cases, the majority report a functional limitation of the hip [5].

Osteoporosis as a whole is an age-related disorder characterized by decreased bone mass. It has been hypothesized that TOP is due to pregnancy as a precipitant of osteoporosis, rather than the cause of the osteoporosis [4,5]. A report by Khastgir, *et al.* reviewed that their patient had pre-pregnancy osteopenia that was exacerbated in the pregnancy state and the degree of bone loss in the patient was similar to bone loss noted in patients who did not have pre-pregnancy osteopenia. Therefore, they believe that the bone loss noted in their patient alone would not have been enough to produce osteoporosis on their own. They concluded that fetal calcium demands induced osteoporosis in the mother [5].

There are two isolated case reports that revealed skeletal changes in the children of patients affected by TOP, but otherwise data on the effects of TOP on the fetus are scarce and subsequently we are unable to establish any direct relationship between TOP and effects on the newborn [2]. Resolution of symptoms occurs naturally postpartum over the course of 6 to 12 months [1-5].

For diagnosis, plain radiographs may demonstrate severe cases of osteopenia, however magnetic resonance imaging is a more sensitive imaging modality that is safe to use in pregnancy that can diagnose skeletal abnormalities like TOP. Management of TOP varies, but the majority of the literature reviews agree on and recommend expectant management [1-6]. Patients are advised to remain non-weight bearing and venous thromboembolism prophylaxis is often recommended. Both physical and occupational therapy is recommended in order to help patients cope with their activities of daily living. Ultimately, because the disease progression is self-limited, surgical intervention is not warranted. In the postpartum period, adjunctive therapy can be considered with bisphosphonates, calcium, and vitamin D [7].

Our patient's pregnancy is complicated by dichorionic/diamniotic twin pregnancy in the setting of a past surgical history of two prior cesarean sections which necessitated a repeat cesarean delivery. However, in the majority of TOP cases, due to the severity of hip pain, Cesarean has become a non-obstetric indication for elective Cesarean for over 70% of reviewed cases [4].

TOP is a rare condition and in order to diagnose early in the clinical course, it is important to recognize the presentation in joints other than the rarely reported TOP of the hip. Diagnosis will allow for the best pregnancy outcomes with reduced maternal complications.

Conclusion

While rarely diagnosed, TOP is more frequently seen in the hip, therefore presentation in the knee is even more rare. The proposed mechanism of disease pathology is unclear, but experts suggest TOP is a multifactorial disease, perhaps influenced by maternal pre-

pregnancy state and bone density. Given the abundance of musculoskeletal complaints in pregnancy, this case highlights the importance of diligently assessing complaints and the utility of diagnostic imaging in the third trimester. Early diagnosis of this disease is essential in preventing further maternal complications. Although TOP is rare, it is important for both obstetricians and orthopedists to include TOP as a differential diagnosis if a pregnant patient presents with substantial pain in their bones.

Declaration of Patient Consent

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

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Nil.

Conflicts of Interest

There are no conflicts of interest.

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