

## Case Report

# Widespread osteonecrosis of long bones following pregnancy

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## Abstract

Osteonecrosis is thought to result from the temporary or permanent loss of the blood supply to the bones. The most common symptom is pain which is localized to the affected condyle. The medial femoral condyle is typically affected, the lateral femoral condyle, medial tibial plateau and patella may also be involved. The purpose of this case report is to highlight the importance of osteonecrosis as a cause of post-partum hip, knee and other joint pain and to review the literature in regards to pregnancy as a possible etiological factor. We presented a case of a 25 year old female who presented to us on 20<sup>th</sup> December 2015 with pain in the right thigh of 10 days duration at the distal half extending up to mid-calf.

## Key words

Osteonecrosis, Long bones, Pregnancy.

## Introduction

Osteonecrosis is thought to result from the temporary or permanent loss of the blood supply to the bones [1, 2]. This disease is also known as avascular necrosis, aseptic necrosis, and ischemic bone necrosis. The femoral head is particularly prone due to its limited collateral circulation, accounting for 20,000 cases per year in the

United States [3, 4]. The average age at presentation ranges from 30 to 50 years with known associations with hyperlipidemia, alcoholism, Cushing's syndrome, hyperuricemia, sickle cell anemia, lupus and rheumatoid arthritis [3-5]. The incidence of Osteonecrosis in the distal femur is second only to that of the femoral head, however, it still represents only 10% of all

cases of avascular necrosis [6]. Pfeifer (1957) reported a case of avascular necrosis of both the femoral and humeral heads immediately following a caesarian section in a woman. Patterson, et al. (1964) reported another case which occurred during post-partum period.

The most common symptom is pain which is localized to the affected condyle. The medial femoral condyle is typically affected, the lateral femoral condyle, medial tibial plateau and patella may also be involved [7-9]. Reddy and Frederick [10] identified a distinct difference between the lateral and medial femoral condyles with respect to both intraosseous and extra osseous blood supply in a cadaveric dye injection study. They hypothesize that this difference may explain the increased frequency with which the medial femoral condyle becomes osteonecrotic in primary osteonecrosis. In secondary osteonecrosis lateral femoral condyle involvement is 60%, is bilateral in 50%, and may be multifocal [11]. Pain may be medial or lateral and is often insidious in onset. Patients may develop symptoms of internal derangement [12]. It is difficult to distinguish between osteonecrosis and other types of intra-articular pathologies on the basis of symptoms. Often unnecessary arthroscopic attempts are applied to these cases. Magnetic Resonance Imaging (MRI) is a sensitive method for the early diagnosis of osteonecrosis. The extension, dimension, signal characteristics, collapses and other accompanying pathologies of a lesion can be clearly identified on MRI [13].

The purpose of this case report is to highlight the importance of osteonecrosis as a cause of post-partum hip, knee and other joint pain and to review the literature in regards to pregnancy as a possible etiological factor.

### **Case report**

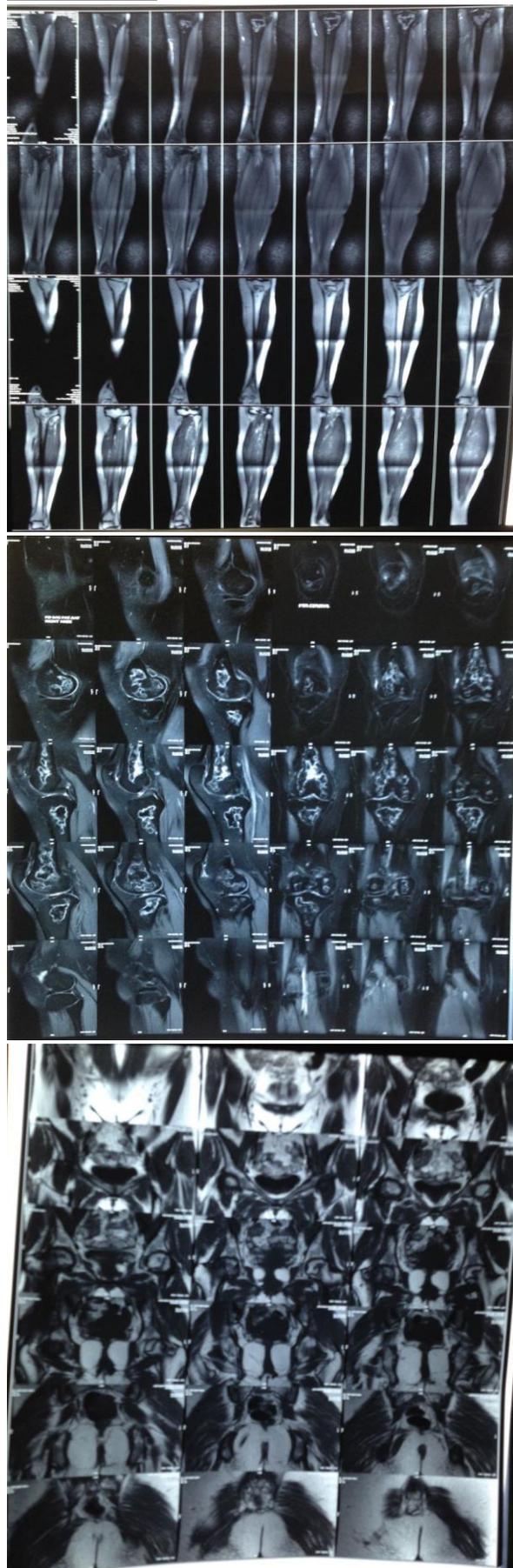
We presented a case of a 25 year old female who presented to us on 20<sup>th</sup> December 2015 with pain in the right thigh of 10 days duration at the distal half extending up to mid-calf.

She was previously seen by us for pain in both hips in November 2014. At that time she was investigated and diagnosed as a case of bilateral avascular necrosis of head of femur of unknown etiology. She underwent bilateral core decompression and was relieved of the pain. Her past history included the delivery of premature baby at 8 months in January 2014. Delivery was normal and conducted at a private hospital. She was given medications to prolong pregnancy to full term which did not happen. One dose of steroid (betamethasone 12 mg) was given just before delivery. The only other history of steroid use was that of a single dose of Dexamethasone 10 mg given in injectable form pre-operatively for a mandible reconstruction surgery 3 years before delivery. Investigation for the pain in distal thigh in the form of X-rays initially did not reveal any abnormality. As the pain did not respond to regular analgesics a MRI of right knee with distal thigh was advised which revealed avascular necrosis of the distal end of femur and proximal end of tibia. This picture mandated a Bone scan which was performed with Technitium99 and revealed avascular necrosis of both humerii, both femoral heads, both femoral condyles and both proximal tibia. Due to the nature of presentation she was advised further investigations which included hemoglobin electrophoresis, sickling, anti cardiolipin antibody, lupus anticoagulant, ANA, pANCA, cANCA, HIV, CBC, ESR, CRP all turned out to be normal (**Photo – 1 to 4**).

**Photo – 1:** X-ray.



**Photo – 2 to 4: MRI.**



In the literature the potential for the steroid to cause osteonecrosis and its format have been varying from single to dual dose. 4000mg of prednisolone equivalence as the threshold for causing multifocal osteonecrosis was suggested [39]. Steroid use in this case has been too small and at 3 years interval does not add up to the suggested levels.

With the above rare presenting features of osteonecrosis with pain at multiple long bone ends a literature search was done which revealed only a few studies which did not have a definitive protocol on management. Papers on the osteonecrosis of the head of femur have given classification and management both medical and surgical depending on the stage of presentation.

A few papers have dealt with the classification and management of the osteonecrosis of the distal femur and proximal tibia. Clinical outcomes with the use of core decompression have been good in patients with Ficat and Arlet stages I and II disease, with a success rate of 79% at a mean follow-up of 7 years [40]. Another study with percutaneous drilling showed a 92% success rate (Knee Society score >80) [41]. Core decompression appears to be a reasonable treatment option for patients with early stage secondary osteonecrosis of the knee with no signs of condylar collapse. Decompression of the distal femur has a better success rates but the overall management of the other sites is not documented.

The number of papers on the osteonecrosis at multiple long bone ends is very few. Through this case report on Osteonecrosis of long bones the common etiological factors like steroid use, alcohol abuse, Thalassaemia, sickling, myeloma etc. were eliminated and pregnancy being the most probable cause in this case was attempted.

**Discussion**

Hip pain during the later stages of pregnancy and during the postpartum period is a common

presentation and is usually related to sciatica, pelvic structural compression and lumbosacral strain [14]. Transient osteoporosis, which is self-limiting and typically resolves within months, and osteonecrosis, which results in femoral head collapse and degenerative changes in the joint, can also cause pain [5, 14, 15]. Transient osteoporosis has been reported in the knee [16], although less frequently than in the hip. It has been noted that the term “bone marrow edema syndrome” (BMES) may be more appropriate, as osteoporosis is not seen on biopsy of this lesion [17]. Some investigators believe Bone marrow edema syndrome represents an early, reversible stage of osteonecrosis [18-21]. The Bone marrow edema syndrome pattern may be more diffuse, however progression to osteonecrosis may be inhibited by abundant new bone formation [17]. Koo and associates [22] suggest that Bone marrow edema syndrome and osteonecrosis may be at opposite ends of a continuum of tissue sensitivity to an ischemic event. If ischemia reaches a certain threshold progression to Avascular necrosis is more likely. Only a few case reports of Bone marrow edema syndrome about the knee exist in the literature [23-32], as such most information on Bone marrow edema syndrome is related to the hip and has been extrapolated to the knee. Bone marrow edema syndrome affects women at one third the rate of men. Affected men are generally middle aged. When women are affected, it is almost exclusively during the third trimester of pregnancy [19]. The lateral femoral condyle is more frequently involved. Treatment is controversial. Some believe that the risk of Bone marrow edema syndrome progressing to Avascular necrosis outweighs the minimal risk of core decompression and recommend performing this procedure more aggressively [19], while others believe the good prognosis of Bone marrow edema syndrome does not justify surgical intervention, reserving core decompression for true Avascular necrosis [33, 34].

The use of MRI is an effective method for evaluating patients with osteonecrosis of the femoral head during pregnancy [14, 15] since the early stages of osteonecrosis are not easily recognized clinically during antenatal and postnatal care [35]. Venous congestion and hypercoagulability are common in the third trimester during pregnancy. There is an increased incidence of thromboembolic phenomenon during pregnancy particularly those who take contraceptive pills. Osteonecrosis is more common on the left side than the right side in pregnant women, which can be explained by the anatomy of venous drainage [5]. The left common iliac vein passes deep to the right common artery and may be subject to excessive compression from the weight of the developing fetus [5, 36].

Other possible etiologies are ovarian hyper stimulation drugs, which have the detrimental effects of hyper viscosity and hyper-coagulability [36, 37] and the mechanical stress or overload by excessive labour and weight gain during the last trimester of pregnancy [14, 15]. Furthermore, many endocrine modifications occur during pregnancy such as parathyroid hyperplasia and the production of estrogen and progesterone by the placenta [15]. These can destabilize endogenous plasma lipoproteins and lipid metabolism in the liver, which could promote fat embolism [5, 14, 36, 38]. Increases in estrogen and progesterone results in increased adrenal cortical activity and levels of adrenal corticosteroids to almost three times the level of a non-pregnant woman [14, 15, 36, 38].

The optimal treatment of osteonecrosis in a young patient is controversial. Conservative methods are usually initiated when there is no evidence of collapse such as restricted weight bearing [5]. Furthermore, many surgical options have been proposed according to the stage of the disease. These include core decompressions, vascularized and non-vascularized grafts, osteotomy and as a last resort option arthroplasty [5, 6, 14].

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