

# Multifocal Avascular Necrosis Complicating Delivery. A Report of two Cases

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## ABSTRACT

*Avascular necrosis (AVN) is characterized by bone death. It affects bones with precarious blood supply. Trauma with disruption of blood supply is a common cause. The aim of this case report is to highlight the difficulty in managing patients with AVN. We present the cases of two young mothers who presented with pains in the shoulder and hip joints. The first is a 22year Para 1+0 housewife referred to our centre with pains in both shoulders and hips of 14 months duration, six months following her last confinement. The Pain was severe, non-radiating, aggravated by movements and relieved with intake of analgesics. She was said to have been in labour for over 24 hours following which she was delivered of a baby boy through spontaneous vertex delivery (SVD). She however had a retained placenta that was removed piecemeal after several attempts at removal failed. The second patient is a 25year old Para 1+0, none alive woman presented with 8years Complaint of pains in both hips with limitation of movements following a difficult delivery 10 months earlier. Both patients were from a low socioeconomic economic background. They were managed with analgesics and physiotherapy and counselled for total joint replacement. Avascular necrosis and osteoarthritis of major joints poses a major challenge in management in a poor resource setting without health financing and insurance.*

**Keywords:** Avascular necrosis, osteoarthritis, hip, shoulder

## INTRODUCTION

Avascular necrosis (AVN) also known as osteonecrosis or ischaemic necrosis affects bones with precarious blood supply and disruption of the vascular supply results in necrosis of the marrow, medullary bone, and cortex<sup>1</sup>. Individual patients developing avascular necrosis usually have more than one risk factor; this indicates that the pathogenesis of AVN is likely multifactorial. Vascular occlusion can occur from trauma involving extra-osseous blood supply as in fracture or dislocation. Altered lipid metabolism can lead to lipid deposition in the femoral head causing femoral hypertension and ischaemia<sup>1,2</sup>. Corticosteroid administration was associated with fat emboli in the femoral heads of rabbits. Other risk factors include intravascular coagulation as seen in familial thrombophilia, hypercholesterolemia, infection, malignancy and pregnancy<sup>3</sup>. Dead bone sets up a process of repair that involves osteoclasts, osteoblasts, histiocytes, and vascular elements. New bone is formed on top of the dead bone, leading to sclerosis that prevents revascularization of the necrotic bone, with resultant abnormal joint remodelling and joint dysfunction.

Ficat and Arlet staged AVN based on radiologic features between early pre-symptomatic stages and late stages where there is collapse and secondary osteoarthritis<sup>4</sup>. Management of early stages will involve use of analgesics and physiotherapy while surgery is indicated once there is structural damage and distortion of the head. We present two cases of radiologically established avascular necrosis and osteoarthritis involving both heads of humerus and femur in order to enrich our knowledge.

## CASE PRESENTATION

### Case 1

We report the case of a 22year Para 1+0 housewife referred to our centre with complaints of pains in both shoulders and hips of 14months duration following her last confinement. There was also history of difficulty in walking of the same duration. The patient problems started after her last delivery when she experienced pains in both shoulders and hips. The pain was severe, non-radiating, aggravated by movements and relieved by intake of analgesics. She was said to be in labour for over 24 hours following which she was delivered of a baby boy through spontaneous vertex delivery (SVD). She however had a retained placenta that was removed piecemeal after several attempts at removal failed. There was much bleeding and patient went into shock

and was resuscitated and transfused with three pints of whole blood. Her pregnancy was booked and she attended ante-natal care regularly and was immunized. Patient is not a known sickle cell disease patient and was confirmed by haemoglobin electrophoresis and has no previous history of hand and foot pain. There was no history of use of contraceptive or prolong steroid. Patient was commenced on analgesics and physiotherapy at the referring hospital and was able to walk with the aid of a walking stick. Examination revealed a young lady, not pale, afebrile and not dehydrated. The respiratory rate was 18 cycles per minute and the chest was clinically clear. The pulse rate was 90 b/min with a blood pressure 100/60mmHg. The abdomen was full and no masses felt. Musculoskeletal examination showed that patient walks with an antalgic gait and the left shoulder was wasted with restriction of movement; active abduction was 0 degree while passive abduction was 20 degrees. The other shoulder and hip joints also were tender during passive movement with restriction of both internal and external rotation movements. She had haematological and radiological investigations with the x-rays confirming Ficat and Arlet stage 3 AVN for the humerus and stage 4 for the hips [Figure 1, 2]. She was counselled on the condition and continued on physiotherapy and possible joint replacement before she was lost to follow up after discharge.



Figure 1. AVN involving both heads of humerus



Figure 2. AVN involving both femoral heads

### Case 2

She was a 25 year P1+0 non alive divorced young lady who presented with an 8years history of pains in both hips. Pain was insidious in onset and is mild to moderate in severity and has gradually been associated with limitation of movements. Pain is relieved by analgesics and worsened by activities. Pain started following her last delivery from a prolonged labour for which she eventually lost the child. She is not a known sickle disease patient and was not on any family planning or steroids. The restriction of hip movements has affected her activities of daily living. Examination revealed a young woman with loss of abduction, adduction, internal and external rotation movement in both hips. Haemoglobin electrophoresis showed genotype AA. Radiographs showed avascular necrosis with secondary osteoarthritis [Figure 3]. She was placed on analgesics while she awaits bilateral total hip replacement.



Figure 3. Osteoarthritis secondary to AVN involving both femoral heads

## DISCUSSION

There are several causes of avascular necrosis of bone and pregnancy related cases are rare<sup>5</sup>. But many cases are classified as idiopathic because of the difficulty in determining the cause. The intraosseous microcirculation is as susceptible to a prothrombotic state as any other part of the circulation<sup>4,5</sup>. Taking Virchow's triad as an example, this may be the result of one or a combination of factors, including endothelial damage, circulatory stasis, or a hypercoagulable state. Endothelial damage may be related to trauma, atherosclerotic lesions, or autoimmune inflammatory connective tissue disorders that may affect the endothelial lining.<sup>5,6</sup> Osteonecrosis of the femoral head is a rare manifestation of pregnancy, especially in healthy women who have no known risk factors for the development of this disorder. The cause of osteonecrosis in these patients is unknown, and the pathogenic mechanism is multifactorial<sup>6,7</sup>.

Different hypotheses have postulated the possibility of amniotic fluid emboli, a relative hypercoagulable state, excessive mechanical strain, and an increase in endogenous steroid production<sup>7,8</sup>. The possibility of amniotic fluid embolism is further enhanced by the difficult delivery with retention of placenta that was removed piecemeal. In pregnancy, the elevated levels of oestrogens and progesterone could induce the development of osteonecrosis by fatty embolism<sup>8</sup>. Pregnancy produces a hypercoagulable state and also causes mechanical stress as a result of the weight gain during pregnancy. Hypercoagulability can cause vascular occlusion and ischaemia and death of bone from thromboembolism<sup>9,10</sup>. To date, the largest case series was described by Montell and associates. In this case series, 13 women developed hip pain late in the second or in the third trimester of their pregnancy. In general, these women tended to have a small body habitus, and during their pregnancy, they had gained excessive weight, indicating that this may have a pathogenic role in the development of osteonecrosis<sup>11</sup>. All the affected women in their series had involvement of their left hip, and four of them had bilateral involvement. Involvement of the both shoulders and hips as seen in our first case is quite rare as most cases reported in the literature involved the hips.

Early diagnosis is important in determining the course of the disease as both patients presented late. The natural history of AVN shows that bone death is an irreversible process leading to secondary osteoarthritis. Management of this condition is quite challenging as both patients are poor with no healthcare financing. The first patient was lost to follow-up while it took the second patient 8 years after developing symptoms to present in hospital. She lacked social support as she was divorced during the course of her illness.

## CONCLUSION

Avascular necrosis should be considered as differential diagnosis of pains involving the large joints during pregnancy and the postpartum period.

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