# Heterotopic Ossification Circumferentia Articularis (HOCA) of Hip Joint: A Case Series and Review of the Literature

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### **Abstract**

**Introduction:** Heterotopic ossification (HO) is often described as the mature, lamellar bone formed in a non-osseous tissue. This new bone formation usually occurs between muscle planes or between muscle and joint capsule. When it surrounds a joint circumferentially, it is known as (HO) Heterotopic Ossification circumferentia articular is circumferential articularis (HOCA). HOCA of the knee has been described earlier in the literature, but this term has never been described for the hip joint in the literature.

**Case Presentation:** We present a case series of two such patients having HOCA of hip joints. In both the cases, there was a history of brain injury and prolonged intubation, which seems the most likely predisposing factors in the causation of HOCA in these hips.

**Conclusion:** We believe that in our patient, the responsible factors for the causation of HOCA could be myriad of factors such as prolonged intensive care unit (ICU) stay with mechanical ventilation and hypoxia, long-standing immobilization, and hypomobility with incomplete flaccid paralysis. The surgical excision is quite challenging, extensive and is not advisable in most cases, and they respond symptomatically to conservative methods including analgesics, bisphosphonates, physiotherapy, and radiation.

Keywords: Heterotopic ossification circumferentia articularis, hip joint, neurological injury,

#### Introduction

When the mature lamellar bone is formed abnormally in the periarticular soft tissue, it is known as heterotopic ossification (HO). Riedel first described HO in 1883 [1]. It is associated commonly with traumatic injuries. There are numerous predisposing factors for the development of HO such as neurological injury either to the brain or the spinal cord, major joint surgery, and burns [2]. However, variation in the incidence of HO and the location of the affected joint is known [3]. According to recent literature, the incidence ranges between 10 and 25% [4]. The incidence of ectopic bone formation or HO following traumatic brain and spinal cord injuries has been found to be between 10-23% and 40-50%, respectively [5]. There are various grades of involvement of the joint by HO, as described by Brooker et al. [6]. We present a case series of two patients with HO circumferentially involving hip joints. This

type of involvement in which a joint is involved circumferentially is known as HOcircumferentia articularis (HOCA).

#### **Case Presentation**

#### Case 1

A 27-year-old male had a road traffic accident and sustained head injury (intracerebral bleed), bilateral shaft femur fractures, compound fracture right patella along with patellar tendon rupture, and facial wounds including laceration of the tarsal plate. Initially, he was resuscitated, followed by locked intramedullary nailing of both femur was done. Post-surgery, his neurological status deteriorated and went into a coma and was ventilated in the intensive care unit (ICU). The fat embolism syndrome was suspected. He also developed acute renal failure on the second post-operative day, for which dialysis was started. In due course of time, he developed chest infection and went

into septicemia. He was put on intravenous broad-spectrum antibiotics. He regained consciousness 3 months later and complained of stiffness in his both hip joints. Both the hips were ankylozed in 65° flexion and adduction. His follow-up X-rays showed a united fracture femur with massive circumferential HO around both hip joints (Fig.1). The heterotopic bone also covered the proximal ends of the nail. A computed tomography scan was done which showed HO involving anterior iliac wings, femoral shafts, and adjacent thigh muscles (Fig. 2 and3). It was decided not to operate, due to severity and diffuseness of the involvement by HO. He was managed by a stat dose of intravenous zoledronic acid (4mg), along with non-steroidal anti-inflammatory drug (NSAID) daily for 6 weeks. His pain gradually reduced with the medical management, but the stiffness persisted.

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Figure 1: A plain radiograph of the pelvis with both hips showing the circumferential presence of diffuse heterotopic ossification, with intramedullary nails in situ.

#### Case 2

A 40-year-male presented with a head injury and intracerebral bleed. He was intubated and admitted to ICU for 4 weeks. Upon recovery, he complained of pain and stiffness in the left hip joint. On examination, he had a severe restriction of movements in left hip. X-ray showed circumferential heterotopic ossification around left hip (Figs. 4 and 5). Due to the extensive and global involvement of the hip by the HO, it was planned to treat it conservatively and not to operate in this case. Intravenous zoledronic acid (4mg) was given stat, along with NSAID daily for 6 weeks. His pain gradually reduced with the medical management but the stiffness persisted.

Both of our cases were managed conservatively, with rehab and medical treatment. Rehabilitation included gentle stretching of the joints and mobilization with a walking frame, with weight bearing as per the patient's pain tolerance. No force was applied in stretching exercises. Transcutaneous electrical nerve stimulation was also used for the pain relief.

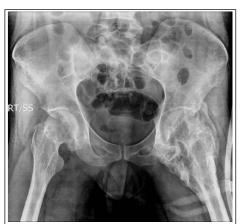


Figure 4: Plain radiograph of the pelvis with both hips showing diffuse heterotopic ossification circumferentially involving the left hip joint.

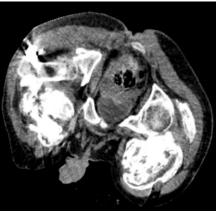


Figure 2: An axial cut of computed tomography showing diffuse heterotopic ossification around both hip joints.

#### Discussion

HO in general is new bone formation in tissues which do not possess properties of bone formation (ossification) and this causes rapid development of calcified bone in soft tissues. It takes only a few weeks for the ectopic bone to develop from immature osteoid [7]. Roberts described in their paper the effects of traumatic brain injury in the pathogenesis of heterotopic ossification where they showed theinvolvement of the elbow in patients with a history of prolonged period of coma and cerebral injuries [2]. Dejerne and Ceiller gave a detailed description of HO after neurological injury. They also in detail discussed the clinical,



Figure 5: Lateral radiograph of the left hip joint showing massive heterotopic ossification.



Figure 3: A reformatted computed tomography scan image of the pelvis and both the femora showing the presence of bilateral interlocking intramedullary femoral nails, with the diffuse bilateral involvement of the hip with heterotopic ossification.

anatomical, and microscopic features of ectopic bone formation in their patient group (soldiers sustaining spinal injuries in the First World War) [8]. There are many known risk factors of HO formation such astrauma, surgery, neurological injury, burn injuries, ankylosing spondylitis, diffuse idiopathic skeletal hyperostosis, and rheumatoid arthritis. [5] (Table 1). Hence, ventilated patients sustaining DAI with prolonged hospitalization are at a higher risk. Other factors include those patients who are genetically predisposed, alterations in acidbase balance which may be explained by decrease in osteoclastic enzyme activity [9]. The process of HO formation begins typically only after a couple of weeks after injury; however, to diagnose this earlier when symptoms have not shown up is seldomly possible. The signs and symptoms range from inflammatory signs around the involved joint along with reduction in range of motion. These may sometimes not present upto as long as 8-10 weeks, and positive radiologically,the lesion of HO is only visible after minimum 1-month post-trauma

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Table 1: Table of risk factors for heterotopic ossification	
Trauma or surgery	Nephrolithiasis
History of previous HO	Hemophilia
Pressure ulcers	Tetanus
Heterotrophic osteoarthrosis	Artificial ventilation
Rheumatoid arthritis	Neurological damage
Osteonecrosis	Genetic predisposition
Hip dysplasia	Burn injuries and Electrocution
Ankylosing spondylitis	Post-traumatic arthritis
Osteoarthritis	Urinary tract infections
Deep vein thrombosis	Sickle cell anemia
GBS	Amputation
Prolonged ICU stay	Diffuse idiopathic skeletal
	hyperostosis
ICU: Intensive care unit, HO: Heterotopic ossification	

episode [10]. HOCA term was first used by Vaishya et al. for the severe case where HO was present all around the knee joint, in a case of Guillain–Barre syndrome [11]. However, the similar involvement of the other joints like hip has not been described, so far. We at this moment present and describe the HOCA of the hip joint, perhaps for the 1sttime. Involvement of hip by HOCA is more debilitating as compared to the knee. Moreover, the incidence of hip involvement is much more common as compared to the

k n e e . The common compounding factor in our cases with that of the described HOCA of the knee by Vaishya et al. [11] includes neurological injury and prolonged intubation and stay in ICU. HOCA can be managed medically, as well as surgically. However, because of the diffuse and circumferential involvement of joint, the surgery is quite challenging, extensive and is not advisable in most cases. Moreover, the reported

outcomes are not very favorable [12]. Drugs such as bisphosphonates, especially etidronate or zoledronic acid, have shown good results [13].NSAIDs have also shown promising results in controlling the pain and the progress of HO formation [14]. Gentle physiotherapy, also, may yield a better functional outcome [5]. Radiotherapy has been reported to help in reducing the size of the swelling [15]. We believe that in our patient, the responsible factors for the

causation of HOCA could bemyriad of factors such as prolonged ICU stay with mechanical ventilation and hypoxia, long-standing immobilization, and hypomobility with incomplete flaccid paralysis.

#### **Conclusions**

The responsible factors for the causation of HOCA could be myriad of factors such as prolonged ICU stay with mechanical ventilation and hypoxia, long-standing immobilization, and hypomobility with incomplete flaccid paralysis. The surgical excision is quite challenging, extensive and is not advisable in most cases, and they respond symptomatically to conservative methods including analgesics, bisphosphonates, physiotherapy, and radiation.

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