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

R Vaishya¹, A Goel¹, A K Agarwal¹, A Vaish¹

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 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 trauma (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 both hip joints. Both the hips were ankylosed 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 and 3). 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.
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.

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 the involvement 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, 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 as atrauma, 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 acid-base 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 up to as long as 8–10 weeks, and positive radiologically the lesion of HO is only visible after minimum 1-month post-trauma.
Heterotrophic osteoarthrosis (HOCA) is much more common as compared to the knee. 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 be myriad of factors such as prolonged ICU stay with mechanical ventilation and hypoxia, long-standing immobilization, and hypomobility with incomplete flaccid paralysis.

### Table 1: Table of risk factors for heterotopic ossification

<table>
<thead>
<tr>
<th>Risk Factor</th>
<th>Description</th>
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<tbody>
<tr>
<td>Trauma or surgery</td>
<td>Nephrolithias</td>
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<tr>
<td>History of previous HO</td>
<td>Hemophilia</td>
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<tr>
<td>Pressure ulcers</td>
<td>Tetanus</td>
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<tr>
<td>Heterotrophic osteoarthrosis</td>
<td>Artificial ventilation</td>
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<tr>
<td>Rheumatoid arthritis</td>
<td>Neurological damage</td>
</tr>
<tr>
<td>Osteonecrosis</td>
<td>Genetic predisposition</td>
</tr>
<tr>
<td>Hip dysplasia</td>
<td>Burn injuries and Electrocution</td>
</tr>
<tr>
<td>Ankylosing spondylitis</td>
<td>Post-traumatic arthritis</td>
</tr>
<tr>
<td>Osteoarthritis</td>
<td>Urinary tract infections</td>
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<tr>
<td>Deep vein thrombosis</td>
<td>Sickle cell anemia</td>
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<tr>
<td>GBS</td>
<td>Amputation</td>
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<tr>
<td>Prolonged ICU stay</td>
<td>Diffuse idiopathic skeletal hyperostosis</td>
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</tbody>
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ICU: Intensive care unit, HO: Heterotopic ossification

### 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.

### References