



## Post-stroke bilateral heterotopic ossification: An acute problem with long-lasting consequences

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Neurogenic heterotopic ossification (HO) is a complication of injury to the central nervous system in which production of lamellar bone within the soft tissues occurs resulting in pain and reduced range of motion (ROM) with eventual ankylosis and loss of functional capacity.<sup>[1]</sup> Heterotopic ossification is frequently seen as a complication of total hip arthroplasty<sup>[2]</sup> and in approximately 20% of traumatic brain injury and spinal cord injury patients.<sup>[3]</sup> Following stroke, HO most commonly involves the hip joint,<sup>[4,5]</sup> occurs on the paretic side and rarely on the non-paretic side.<sup>[6,7]</sup> Bilateral HO following stroke is even rarer with the most recent cross-sectional study identifying bilateral HO in only 0.2% of all stroke patients over a 10 year period.<sup>[8]</sup> In this article, we present a case of extensive established bilateral HO of the hips occurring shortly after the onset of stroke.

### CASE REPORT

A 77-year-old female patient with a five-month history of stroke, right-sided weakness and left knee contracture presented to our Physical Medicine and Rehabilitation Department. Following the onset of stroke, the patient was initially treated in an intensive

### ABSTRACT

Heterotopic ossification (HO) is a complication of injury to the central nervous system in which production of lamellar bone within the soft tissues occurs resulting in pain, reduced range of motion (ROM) and loss of functional capacity. Heterotopic ossification is rarely seen in stroke patients and mostly affects the paretic side. In this article, we present a case of established bilateral HO of the hips soon after stroke onset. A 77-year-old female patient with a five-month history of stroke presented to our rehabilitation clinic. Physical examination revealed sensorimotor aphasia, right-sided hemiplegia, bilateral painful limited ROM of the hips and left knee contracture. An anteroposterior X-ray of the pelvis revealed previously undiagnosed bilateral HO of the hips. The patient and her relatives declined operative interventions. The patient was discussed in the departmental meeting and it was agreed that she would not be able to partake in an active inpatient rehabilitation program. She was discharged with a home exercise plan. This case highlights the importance of HO, of both the paretic and non-paretic side, being included in the differential diagnosis of post-stroke patients presenting with joint pain and reduced ROM, both acutely and in the long-term. This may aid the timely diagnosis and management of HO, a pathology which has detrimental effects on functionality.

**Keywords:** Bilateral, hemiplegia, heterotopic, hip, ossification, stroke.

care unit (ICU) before being transferred to the neurology ward. Following discharge from the ward, a monoarthritis of the left knee developed. According to the patient's relatives, the patient's knee was evaluated by an orthopedics and traumatology team; however, no details of the evaluation/investigations/treatment applied are known. To date, the patient had not partaken in a home exercise program or received physical therapy. The patient's past medical history included hypertension, atrial fibrillation, heart failure and a right total knee replacement five years previously due to osteoarthritis. Prior to development of the stroke, the patient was independent of activities of daily living.

Physical examination revealed a Glasgow Coma Score of 12 with motor and sensory aphasia. There

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**FIGURE 1.** Bilateral heterotopic ossification of hips.

was a flaccid paralysis of the right side. No spasticity was detected. The patient was incontinent of urine and feces. Examination of the musculoskeletal system revealed bilateral painful and limited ROM of the hips and left knee contracture. There was no effusion of the joints. The patient's Functional Independence Measure<sup>[9]</sup> score was 23.

The patient's initial routine blood tests were normal. The erythrocyte sedimentation rate was 69 mm/hour and the C-reactive protein was 11.4 mg/L. An anteroposterior X-ray of the

pelvis revealed extensive bilateral periarticular HO (Figure 1). A two-way bilateral X-ray of the knees revealed a total knee replacement on the right side and extensive osteoarthritis of the left knee (Figure 2). Pharmacological treatment with indomethacin was commenced. The Orthopedics and Traumatology Department was consulted for further management of the bilateral hip HO and left knee contracture. Initially, arthroscopic debridement of the left knee was recommended; however, the patient's relatives did not consent to the procedure. Due to the patient's limited cooperation, immobility, poor right-sided motor recovery, bilateral limited hip movement and left knee contracture, it was decided that she would gain little benefit from an active inpatient rehabilitation program. The patient was discharged with appropriate analgesia and a home exercise plan with a view to follow-up in the outpatient clinic in a month's time.

## DISCUSSION

Bilateral HO occurring soon after stroke is a subject which remains scarcely covered in the literature.<sup>[10,11]</sup> A recent cross-sectional study by Cunha et al.<sup>[8]</sup> of stroke patients admitted to seven rehabilitation centers over a period of 10 years found an annual prevalence of HO of 1.3%.<sup>[8]</sup> Moreover, only 0.2% of the cases identified had bilateral HO.

In this case, the early diagnosis of HO of the hips was missed. The early stages of HO and resultant hip



**FIGURE 2.** Osteoarthritis and varus deformity of left knee joint.

pain may not have been detected due to the patient's poor cognitive function and immobility. For the affected patient, it is crucial that this condition is recognized and diagnosed early so that ankylosis can be prevented by timely treatment.<sup>[12]</sup> The fact that there was bilateral established HO by the time the diagnosis was established underlines the fact that awareness on this matter requires further attention, particularly in bed-bound patients in which hip examination may be more challenging. Often, patients initially experience pain and inflammation of the affected joint; therefore, differential diagnoses such as spasticity, monoarthritis and cellulitis may primarily come to mind resulting in a diagnosis of HO being overlooked.<sup>[13,14]</sup>

Moreover, complications associated with HO such as urinary tract infections, deep vein thrombosis and overlying pressure ulceration<sup>[15,16]</sup> deem early and correct management imperative.

Management of HO is often targeted at preventing the condition from occurring in unaffected patients. Single-dose radiotherapy or oral nonsteroidal anti-inflammatory drugs such as indometacin and maintaining joint ROM post-stroke are used as HO prophylaxis and treatment.<sup>[17]</sup> Preliminary studies and case reports on the role of extracorporeal shock wave therapy (ESWT) in the treatment of neurogenic HO following traumatic brain injury and spinal cord injury suggest that ESWT may reduce pain and increase ROM of the affected joints,<sup>[18-20]</sup> however, excisional surgery remains the treatment of choice to restore joint mobility.<sup>[6,21]</sup> In this case, the contracture of the osteoarthritic left knee may have developed secondary to the neurogenic HO of the hips, as this results in reduced mobility and poor positioning of the patient within bed. Therefore, surgical intervention to the left knee was initially offered to the patient.

Causative factors for the development of HO remain to be elucidated. Recent studies of stroke patients have concluded that the presence of hemorrhagic stroke/ischemic stroke with hemorrhagic transformation, severity of the initial neurological lesion, presence of spasticity and cognitive impairment are risk factors for the development of HO.<sup>[5,7,8,22]</sup> Cases of severe HO, such as the case reported here, involve two or more joints and have been associated with ICU stay and the use of invasive ventilation in the acute phase of stroke.<sup>[8]</sup>

This case describes a patient in which the diagnosis of bilateral HO of the hips was delayed and highlights the importance of HO being included in the differential diagnosis of post-stroke patients

presenting to the Orthopedics Department with joint pain, swelling and reduced ROM both acutely and in the long-term following stroke. Equally, although rare, this case reiterates that development of HO in the non-paretic side following stroke must be kept in mind to provide timely management of a pathology which is difficult to treat, and can result in ankylosis, to avoid lasting detrimental effects on patient functionality.

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