

Functional Outcomes of the Surgery and Rehabilitation in a Challenging Case of Heterotopic Ossification after Encephalitis

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ABSTRACT

Heterotopic ossification is the formation of the lamellar bone where normally osseous tissue does not exist. Since heterotopic ossification can cause severe functional loss, it is a challenging condition for both clinicians and patients. Neurogenic heterotopic ossification is a rare condition after encephalitis. Likewise, in this paper, we have presented a challenging case of heterotopic ossification after viral encephalitis and functional outcomes after the management of heterotopic ossification.

Keywords: Heterotopic ossification, hip, loss of function, meningitis, rehabilitation

Resultados Funcionales de la Cirugía y Rehabilitación en un Caso Difícil de Osificación Heterotópica Luego de una Encefalitis

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RESUMEN

La osificación heterotópica es la formación de hueso laminar donde no existe tejido óseo normalmente. Como que puede causar severas pérdidas funcionales, la osificación heterotópica es una condición difícil para médicos y pacientes. Es también una condición rara después de la encefalitis. Asimismo, en este trabajo presentamos el reto de un caso de osificación heterotópica después de una encefalitis viral y los resultados funcionales después del tratamiento de la osificación heterotópica.

Palabras claves: Osificación heterotópica, cadera, pérdida de función, meningitis, rehabilitación

West Indian Med J 2015; 64 (3): 283

INTRODUCTION

Heterotopic ossification (HO) can be defined as the formation of lamellar bone where normally osseous tissue does not exist. While neurogenic HO has been reported as a frequent complication after traumatic brain injury and spinal cord injury, it has been rarely reported after stroke, multiple sclerosis, arachnoiditis, epidural abscess and tabes dorsalis (1). On the other hand, HO development after encephalitis has been reported only a few times in the literature (2–6). To the best of our knowledge, surgical and rehabilitation outcomes of HO after encephalitis have been reported only in three reports (2, 4, 5). Nonetheless, there are limited data for the risk factors, rehabilitation programme, and details of the functional outcomes

after surgery in relevant documents. We have reported a challenging case of HO after viral encephalitis and functional outcomes after the management of HO.

CASE REPORT

A 27-year old man was seen due to decreased range of motion (ROM) in his lower limbs and severe functional loss. In his medical history, he had been diagnosed with viral encephalitis 10 months ago. He had stayed in a coma and had been intubated for 30 days, and had been followed-up in the intensive care unit for 45 days. Although combinations of different anti-epileptic drugs were used, he had generalized tonic-clonic seizures lasting 20–30 seconds twice a week during that period. He had surgery for a pressure ulcer in his sacrum and had deep vein thrombosis in the right lower limb as well.

On physical examination, both hips joints were fixed in abduction and external rotation (Fig. 1A).

Range of motion of the right knee joint was between 45 and 60 degrees, and that of the left between 30 and 90 degrees. The Standardized Mini Mental Test Score was 30/30. Lower

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Fig. 1: Picture demonstrating functional status and contractures of the patient before management (A) and after management (B).

limb motor muscle test, tonus evaluation and deep tendon reflexes could not be performed due to contractures. He had no sensory deficits. According to Functional Independence Measure (FIM) Motor Scale, he was completely dependent in his activities of daily living (24/91). He was not able to sit due to contractures. He could merely lie in a supine or prone position. Although he had no incontinence, he could not be transferred to the bathroom.

Concerning the laboratory investigations, alkaline phosphatase (ALP) level was 239 u/L (n: 38–155 u/L). Complete blood count, erythrocyte sedimentation rate (ESR), plasma calcium and phosphate levels were all within normal limits. Pelvis radiograph demonstrated HO around both hip joints and femoral necks (Fig. 2).



Fig. 2: Anterior-posterior pelvis radiograph showing heterotopic ossification around bilateral hip joints and femoral necks.

Pulmonary function tests were consistent with reduced endurance of respiratory muscles. Breathing exercises and inspiratory muscle training and positive expiratory pressure devices were recommended. He could not attend an upper extremity ergometer programme due to the function loss. On consultation with neurology department, the seizures were treated with a three anti-epileptic drug combination (zonisamide, lamotrigine and sodium valproate). The patient was referred to the orthopaedics department. After HO was assessed as mature by three-phase bone scan, the patient underwent surgical excision of the ossification tissue. The surgery comprised bilateral tenotomies of the sartorius, rectus femoris and iliopsoas tendons, and right iliotibial band release. Both hip and knee joints were manipulated to check the adequacy of the surgery (Fig. 3).

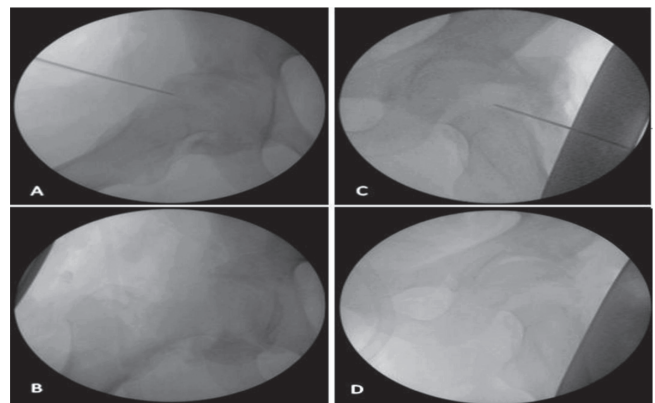


Fig. 3: Preoperative right (A), left (C) and postoperative right (B) and left (D) scoping images.

After the surgery, the patient was transferred to the rehabilitation clinic. Indomethacin 100 mg/day for four weeks was given for prophylaxis. Plastic knee-ankle-foot orthosis was used to provide proper positioning. Gentle range of motion exercises, balance-coordination, posture and strengthening exercises were applied. After regaining the sitting balance, the patient attended the upper extremity ergometer programme. The patient was verticalized within the parallel bars, and thereafter walking training was applied. After a four-month rehabilitation programme, the muscle strength was 5/5 except the right ankle muscles. Ankle-foot orthosis with metal bars was given for the right side. Overall, the patient was able to walk with an ankle-foot orthosis and a tripod assist (Fig. 1B). There was also marked improvement in FIM Motor Scale (73/91).

DISCUSSION

Heterotopic ossification can present with pain, swelling, warmth and decrease in ROM. Further, it can result in nerve and vascular insult, ankylosis and severe function loss depending upon its location (1). Concerning HO localization after encephalitis, most cases have been reported around the

hips and rarely around the knee and elbow (2–6). Infection, deep vein thrombosis, fracture, complex regional pain syndrome and septic arthritis should be taken into account for the differential diagnosis (1).

The exact aetiopathogenesis is not clearly known, yet the three following factors have been reported to be required for new bone formation: stimulating factor, an osteoprogenitor mesenchymal stem cell, and a proper environment for osteogenesis (7). In our patient, a neurologic insult occurrence, hypoxia and endotracheal intubation, prolonged coma and a period of pressure immobility ulcer and its surgery, epileptic seizures, deep vein thrombosis and being late for a comprehensive rehabilitation programme were the stimulating and risk factors for HO. Moreover, generalized tonic-clonic seizures might stimulate HO. Likewise, Tay and Lim (4) reported a case with intractable seizures after encephalitis, whereby seizures might have caused trauma and hypoxia. To eliminate these risk factors, the seizures were treated with aggressive anti-epileptic combinations and the patient was mobilized as soon as possible after the surgery. If the patient had received early rehabilitation when he had been diagnosed with encephalitis, HO might have not developed. Afterwards, HO made the clinical circumstance and its rehabilitation more challenging.

With respect to the laboratory investigations in HO, ALP and creatinine phosphokinase can be helpful. In addition, creatinine phosphokinase is correlated with the severity of HO.

Three-phase bone scan is the most sensitive imaging tool to detect early HO; however, it has low specificity. Radiographs are quite sensitive (7). Ultrasound is also a convenient imaging tool to detect early HO, whereby the “zone phenomenon” is the specific sign of ultrasound (8). Continuous passive motion devices, indomethacin and other non-steroidal anti-inflammatory drugs, radiotherapy and early rehabilitation have been reported to be effective in preventing HO in different sample groups (9–12).

Since HO is a rare condition after encephalitis, the data for the treatment belong to the population with traumatic brain and spinal cord injuries. While there is a high level of evidence for medical options in the spinal cord injury population, there is a high level of evidence for surgical data in the traumatic brain injury population (13). On the other hand, timing for surgery is controversial. The classical approach for the surgical excision of HO suggests surgery after maturation [12 to

18 months] (13). On the contrary, there are also studies suggesting early excision (14). In addition, excision of HO makes the rehabilitation programme more effective and easier (13, 14).

In short, although effective, the surgical excision of HO is a challenging method with a high risk of infection, bleeding, nerve and vascular insults. Surgery and rehabilitation are the two adjunctive treatment options, and they are vital for improving activities of daily living and functional activity regain.

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