



Case Report

Neurogenic heterotopic ossification causing sciatic nerve entrapment in children: A case report.

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Abstract

Heterotopic ossification (HO) is an abnormal formation of mature lamellar bone in soft tissues. It is usually containing bone marrow. Neurogenic HO is rare in pediatric practice and occurs after insult to the nervous system. We herein present one of the first reports of sciatic nerve entrapment caused by neurogenic HO in a 4-year-old boy presenting with severe traumatic brain injury. The aim was to highlight clinical and radiological characteristics and to discuss the most accurate diagnostic tools.

Keywords

heterotopic ossification; children, sciatic nerve; entrapment.

Introduction

Neurogenic HO is seen after central and less frequently peripheral nervous system insults, including spinal trauma and traumatic brain injuries (TBI). The pathogenesis may depend on the transformation of mesenchymal to bone-forming cells in response to a variety of stimuli. Reports suggest that stimuli may be due to immobilization, muscles microtrauma, spasticity of special muscle groups, disturbance of the protein and electrolyte balance, alteration of vasomotor outflow, and tissue hypoxia [1,2].

The prevalence of HO after spinal cord injuries is estimated at 20-25% and following traumatic brain injury (TBI) at 10-23% (5) in adult population. It is less frequent among children, estimated at 10% after spinal cord injury (SCI) and TBI [3]. This entity remained almost unreported in children and might be the misdiagnosed cause of handicap in several cases.

Observation

A 4-yr-old boy presented with severe TBI after a domestic accident. Initial management was marked by long coma and several neurosurgical procedures. He was admitted to the rehabilitation unit five months after the accident. He had cranial fracture and cerebral contusion complicated with infection, diffuse axonal injury and cerebellar contusion. On admission the patient was aphasic. He had spastic flexion deformities in upper limbs and spastic extension deformities in lower limbs. Elbow, hip, knee and ankle joint ranges of motions were limited bilaterally. Substantial neurological examination could not be performed because of joint limitations, but gross motor strengths were tested using medical research council scale (MRC) revealing 2 on the right and 3 on the left side. He had action tremor and static balance dysfunction. Functional independence measure for children (weeFIM) score was 22, careful goniometric measurement of the hip showed bilateral limitation in flexion (60 on the right side and 80 on the left side), rotations were completely impossible on the right side and severely limited on the left side. The patient showed signs of suffering upon mobilization of the hip. Neurogenic HO was suspected. Standard radiography showed bilateral islands of calcification around the hip Brooker grade 1 (figure 1). Alkaline phosphatase in the blood serum were as high as 3 times the normal rate (348 U per liter). Computed Tomography of the hips showed posterior ossification in the coxofemoral joints consistent with mature neurogenic ossification surrounding the sciatic nerve on both sides (figure 2,3). Electrodiagnostic evaluations were consistent with axonal degeneration of the right sciatic nerve. The rest of the muscles, including the quadriceps, gluteus Maximus, gluteus medius, and lumbar paravertebral muscles, were normal. Rehabilitation program was carried on with intensive range-of-motion exercises for the elbow, hip, knee, and ankle joints. Paracetamol was used for pain treatment. He was not proposed for a surgical treatment, because the sciatic nerve entrapment was not responsible for the spastic gait disorder. we decided to start with botulinum toxin injection to the gastrocnemius, soleus and popliteus to straighten the leg and the foot.



Figure 1: plain radiography showing bilateral islands of calcification around the hip (Brooker grade 1).



Figure 2,3: CT scan of the hips showing posterior heterotopic ossification causing compression to the sciatic nerve.

Discussion

Heterotopic ossification (HO) is defined as the extraskeletal bone formation in soft tissues, blood vessels, ligaments and muscles. The pathogenesis is still unclear and involves multiple complex inflammatory cascades mediated by different neuroinflammatory substances. Three types of HO have been described: traumatic, neurogenic and hereditary [4]. The disease always appears caudally to the lesion level, mainly in the hip, knee, elbow and shoulder joints. The hip is the most affected joint in children [5]. A sciatic nerve lesion secondary to heterotopic ossification compression is rare even in adult population. Release of the entrapped sciatic nerve may restore functions partially or completely. However, disabilities are frequently permanent due to late diagnosis, autonomic dysreflexia and muscle spasticity. An early electrophysiologic study maybe useful to determine the location and severity of nerve damage in severe SCI and TBI cases [6]. Prevention of HO for high-risk injury patterns may reduce the morbidity rate and enhance the functional prognosis. Susceptibility for HO formation is a diverse process with several leading factors still to be defined. Steroidal and non-steroidal anti-inflammatory as well as bisphosphonate and radiation therapy are routinely used to modulate inflammation in high-risk patients. Once established, only surgical excision of HO neoformations with release of nervous adjacent structures can ensure functional improvement [7].

Conflict of Interest: None

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