Heterotopic Ossification in Patient with Spinal Cord Compression due to Thoracic Spinal Actinomycosis: A Very Rare Case Report

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ABSTRACT

Introduction: Heterotopic ossification is common in patients with traumatic brain injury, spinal cord injury and after arthroplasty, however, it is rarely seen in paraplegic patients with spinal infection especially spinal actinomycosis. Method: We report a case of spinal cord compression caused by Actinomyces israelii, complicated with heterotopic ossification (HO) in both hip joints in 42 year-old Malay gentle man. He presented with gradual onset of bilateral lower limb weakness associated with constitutional symptoms and difficulty in passing urine for 3 months. On physical examination, his neurological level was T12 AIS (American Spinal Injury Association Impairment Scale) B. Bony swelling was noted over antero-lateral aspect of the proximal thigh (Left). The limitation of range of motion in both hip joints was noted. Results: Laboratory tests indicated leukocytosis with raised ESR and C Reactive Protein. Alkaline phosphatase was 176 IU/L. Radiograph of the pelvis (AP view) showed heterotopic ossification in both hip joints and it was graded as Class 4 on Brooker's classification. Magnetic Resonance Imaging (MRI) scan of the thorax showed paraspinal mass at thoracic (T2-9) region which extended intraspinally causing compression of the spinal cord. CT guided biopsy was done and HPE confirmed actinomyces infection. Therefore, decompression and spinal fixation was offered but patient refused and opted for conservative management. Conclusion: Although heterotopic ossification is rarely seen in patient with spinal cord compression due to thoracic spinal actinomycosis, physicians should bear in mind this rare incidence so as to get early diagnosis and give prompt and effective treatment.

KEY WORDS

spinal actinomycosis, spinal cord compression, heterotopic ossification, Actinomyces israelii

INTRODUCTION

Heterotopic ossification is common in patients with traumatic brain injury, spinal cord injury and after arthroplasty, however, it is rarely seen in paraplegic patients with spine infection especially spinal actinomycosis. Spinal cord compression due to actinomycosis is unusual and seen only in less than 5% of infections1. None has been reported on heterotopic ossification in spinal actinomycosis so far.

CASE REPORT

We report a case of spinal cord compression caused by Actinomyces israelii and complicated with heterotopic ossification (HO) in both hip joints in 42 year-old Malay gentle man. He presented with gradual onset of bilateral lower limb weakness associated with constitutional symptoms and difficulty in passing urine for a duration of 3 months. Upon physical examination, his neurological level was T12 AIS (American Spinal Injury Association Impairment Scale) B. Bony swelling was noted over the antero-lateral aspect of proximal thigh (L). The limitation of range of motion in both hip joints was noted.

RESULTS

Full blood count showed leukocytosis - 27.9 x 10^9/L (4.0-10.0), predominantly neutrophils -25.3 x 10^9/L (2.0 -7.0), Haemoglobin level was 8.5 g%, ESR was114 mm/1^st hr. Albumin was 20 g/L, Alkaline phosphatase was 176 IU/L and C Reactive Protein (CRP) was 18.14 mg/dl. Magnetic Resonance Imaging (MRI) scan of thorax displayed paraspinal mass at thoracic (T2-9) region (Figure 1,2). There was also an intraspinal extension of the lesion with compression on to the spinal cord.
HO occurs in patients with spinal cord and brain injury, major joint surgery and burns. In 1918, Dejerne and Ceiller first diagnosed heterotopic ossification after a case of neurological injury. They made the discovery of ectopic bone formation in a soldier who had sustained spinal injuries in the First World War\(^6\). Patients with conditions of sickle cell anemia, hemophilia, tetanus, poliomyelitis, multiple sclerosis, and toxic epidermal necrolysis and those suffering from burn have also been diagnosed with HO\(^7\). Also, there are cases of idiopathic HO which has occurred in difference to a recognized precipitating condition. Ohnmar et al reported a case of massive heterotopic ossification in Guillain-Barré syndrome with the underlying possible pathology of prolonged ICU stay on mechanical ventilation and hypoxia, long-standing immobilization and hypomobility\(^8\).

Highly controversial, the pathophysiology of heterotopic ossification is still on debate. Amongst the aetiologies are cases such as inflammatory factors in relation to denervated tissues, disruption in calcium homeostasis, immobilisation, long-term pressure on periartricular structures, microtrauma, vascular stasis, hypoxia, hyperthermia, and in due cases, genetic factors aetiologies\(^9\).

Although there are many mechanisms involved in the formation of HO after neuro-trauma, none of the articles have highlighted the possible mechanisms of HO in spine infection especially spinal actinomycosis. Spinal cord compression due to actinomycosis is unusual\(^6\) and seen only in less than 5% of the spine infections\(^1\). Only limited numbers of similar cases have been reported so far.

Actinomycosis is a chronic, suppurative granulomatous infection which is usually caused by Actinomyces israelii, an anaerobic, saprophytic bacteria\(^7\). In this case, patient developed thoracic spinal cord compression due to actinomycosis. His neurological deficit was T12 AIS B and complicated with heterotopic ossification in both hip joints. This is a very rare case of heterotopic ossification in spinal actinomycosis. Incidence of HO in non-traumatic SCI was 4.4%\(^8\). No similar case has been reported so far.

Multi-factorial aetiology with several risk factors is associated with HO. Genetic predisposition should be considered as part of the aetiology in developing HO since it does not develop in all patients with head injuries\(^6\).

A pivotal role is played by the pluripotent progenitor cells which are Mesenchymal stem cells or human bone marrow stromal stem cells in the development of heterotopic ossification. Existing in post-natal condition, they exhibit the characteristics of stem cells, portraying the ability to generate cartilage, bone, muscle, tendon, ligament and fat\(^10\).

There are several stimulating factors in human body such as Bone morphogenetic protein (BMPs), interleukin-1β, Growth hormone, prolactin, insulin-like growth factor type-I and basic fibroblast growth factor. BMPs together with the correct local environment could cause the transition of mesenchymal stem cells to bone resulting in the formation of HO after head injury\(^11\).

Other local tissue environment that favors the formation of HO is microvasculature changes such as alterations in the endothelial cells, basement membrane of capillaries, and small vessels, changes in oxygen tension, pH and blood flow\(^12\).

In animal models, subperiosteal lamellar bone formation was enhanced by prostaglandin E2 (PGE2)\(^13\). PGE2 may be an inducer of bone formation in human. The 24-hour PGE2 urinary excretion has been shown to have increased in patients with acute spinal cord injury (SCI) who developed heterotopic ossification\(^14\). Excretion of PGE2 continues until the bone reached a level of maturity. The clinical signs and symp-
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Heterotopic Ossification (HO) could occur due to myriad factor, in particular to this case, a result of inflammatory factors consequential from denervated tissues and infection, calcium homoeostasis disruption, prolonged pressure in effect to poor bed mobility and body-positioning, microtrauma as well as poor transferring techniques. This case reports highlights the possibility of occurrence of HO in thoracic actinomycosis although it is very unusual.

REFERENCES