CASE REPORT

Idiopathic heterotopic ossification in the intensive care setting

J E Lane, R J Dean, G D Foulkes, P W Chandler

Heterotopic ossification is characterised by the periarticular deposition of ectopic bone. It typically occurs after trauma, neurogenic injury, or congenital causes. Idiopathic heterotopic ossification has been rarely reported. A patient who developed idiopathic heterotopic ossification in the intensive care unit without any known predisposing conditions is presented.

Heterotopic ossification is the formation of mature lamellar bone in periarticular soft tissue and is often associated with traumatic injuries.\(^1\)\(^2\) It was first described by Riedel in 1883.\(^1\) Heterotopic ossification is subclassified as traumatic myositis ossificans, neurogenic heterotopic ossification (myositis ossificans circumscripta), and myositis ossificans progressiva.\(^3\)\(^4\) It typically occurs after closed head injury, spinal cord injury, and thermal burns but has also been described with neuromuscular blockade, trauma, acute polymyelitis, and encephalitis.\(^5\)\(^6\)

The pathophysiology of heterotopic ossification is unknown. Several theories have been proposed, including inflammatory factors derived from denervated tissues, disrupted calcium homoeostasis, immobilisation, prolonged pressure on periarticular structures, microtrauma, vascular stasis, hypoxia, hyperthermia, and genetic factors.\(^7\)\(^8\) Heterotopic ossification characteristically begins approximately two weeks after injury; however, diagnosis is often delayed.\(^9\) Common signs and symptoms include decreased range of motion, pain, swelling, and erythema. These non-specific clinical signs may not appear until eight to 10 weeks after the inducing injury.\(^1\) Positive radiographic findings may not appear for four weeks.\(^2\) An acute rise in serum alkaline phosphatase and a transient depression in serum calcium may occur within the first two weeks. However, these are non-specific laboratory findings and may not be helpful in early diagnosis.\(^4\) The determining characteristics of heterotopic ossification include a periarticular location, an intact cortex, a lucent zone between cortex and ossification (“string sign”), peripheral density of calcification, and contraction of the ossification zone with maturity.\(^8\) A rise in blood alkaline phosphatase, hydroxyprolinuria, and serum creatine phosphokinase have been reported, while calcium and phosphorus typically remain within normal limits.\(^2\)\(^8\)

Treatment of heterotopic ossification is controversial and includes the use of bisphosphonates, non-steroidal anti-inflammatory agents, prophylactic irradiation, and surgical excision.

The occurrence of idiopathic heterotopic ossification in critically ill patients in the absence of predisposing conditions has been rarely reported.\(^9\) We describe a patient who developed idiopathic heterotopic ossification in the intensive care unit with no known predisposing injuries or conditions.

CASE REPORT

A 38 year old African-American woman was admitted to hospital with complaints of headaches, sweats, chills, productive cough with dyspnoea, and fever. Her past medical history included hypertension and an uncomplicated childbirth. She denied the use of tobacco and alcohol, and had no allergies. Her medications included ranitidine, sertraline, and atenolol.

Physical examination revealed an acutely ill young woman who was alert and oriented. Vital signs included the following: blood pressure 98/70 mm Hg; pulse 166 beats/min; respirations 28 breaths/min; temperature 39.1°C. Pulmonary examination revealed bilateral crackles throughout all lung fields. Cardiovascular examination demonstrated resting tachycardia without murmurs or bruits. Her abdomen was mildly protuberant with active bowel sounds. Tenderness and hepatomegaly were noted over the hepatic capsular area. Neurological examination was normal.

Laboratory evaluation disclosed the following (reference range in parentheses): total creatine phosphokinase 1531 U/l (26–140 U/l); prothrombin time 12.9 sec (10.9–13.1 sec); partial prothrombin time 36 sec (25–35 sec); serum lactate dehydrogenase 882 U/l (208–378 U/l). Haematological analysis revealed a white blood count of 8900 \(\times\) 10\(^6\) (4.5–11.0 \(\times\) 10\(^6\)) with 92% blasts, a platelet count of 87 \(\times\) 10\(^6\) (150–450 \(\times\) 10\(^6\)), and a haemoglobin of 96 g/l (120–160 g/l). Serum potassium was decreased to 2.7 mmol/l (3.5–5.1 mmol/l). Arterial blood gas analysis included the following: pH 7.52; carbon dioxide pressure 4.0 kPa; oxygen pressure 5.9 kPa (room air). A chest radiograph revealed changes consistent with bilateral interstitial pneumonia.

The patient was transferred to the intensive care unit for progressive hypoxic respiratory failure and placed on mechanical ventilation. She required prolonged ventilatory support secondary to acute respiratory distress syndrome and bilateral pneumonitis. She continued to remain febrile and neutropenic. Transoesophageal echocardiography demonstrated a broad based sessile vegetation on the mitral valve. She was noted to have a haematoma of the mitral valve suspicious of bacterial endocarditis. A diagnosis of leukaemia was made from the peripheral smear. She was treated with a number of antimicrobial agents, including vancomycin, amphotericin, and ciprofloxacin, for presumed endocarditis.

Her platelet count continued to decrease and she developed joint stiffness in her elbows and knees. She developed decreased range of motion and pain with subsequent diffuse muscle wasting of the upper extremities. Heterotopic ossification was diagnosed approximately six weeks after initial admission based on both laboratory and radiographic findings. Radiographs showed striking zones of heterotopic ossification of the periarticular regions at knee and elbow joints (figs 1 and 2). Alkaline phosphatase reached a peak of 903 U/l (42–98 U/l).

DISCUSSION

Idiopathic heterotopic ossification has been rarely reported.\(^9\) Heterotopic ossification is typically categorised as due to
Heterotopic ossification is characterised by periarticular deposition of ectopic bone. Heterotopic ossification typically occurs after trauma, neurogenic injury, or congenital causes. The pathophysiology is unknown. Common signs and symptoms include decreased range of motion, pain, swelling, and erythema. Laboratory findings are non-specific and may not aid in early diagnosis.

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