

Traumatic Heterotopic Ossification of the Elbow in Children – a Case Report

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Abstract. Heterotopic ossification is defined as the formation of mature, lamellar bone within soft tissues other than the periosteum. We present a case of a 9-year-old girl with formation of heterotopic bone tissue in the elbow after surgical treatment of a fracture of the lateral humeral condyle. Eight months after the first operation the girl was re-operated to resect the heterotopic bone tissue. We believe that radical surgical excision of heterotopic bone, following physiotherapy around the elbow in children can improve the function of the extremity.

Introduction

First documented in 1692 by Guy Patin, heterotopic ossification is defined as the formation of mature, lamellar bone within soft tissues other than the periosteum. This pathologic process may occur in tissue such as skin, subcutaneous tissue, skeletal muscle and peri-articular tissue (1). The historical terms for heterotopic ossification have been superseded, but ectopic ossification and myositis ossificans are used interchangeably with the term heterotopic ossification (2). Three types of heterotopic ossification have been described: traumatic – following operative procedures, fractures or dislocations, neurogenic – occurring after closed head injuries, insults to the spinal cord, or central nervous system infections, and genetic (myositis ossificans progressiva – a rare, autosomal dominant disease whereby skeletal muscle ossifies) (1-4). An increased incidence of heterotopic ossification has been found in persons over the age of 30. The incidence of heterotopic ossification in children appears to be lower than that in adults (8-22.5%) (5). The aetiology of traumatic heterotopic ossification remains uncertain. During the past 50 years, a number of theories have been developed. The inappropriate differentiation of pluripotential mesenchymal cells into osteoblastic stem cells has been suggested as a potential cause of osteogenesis in connective tissue (3, 4). In the literature there is very little about heterotopic ossification treatment in children. We describe the case of a 9-year-old girl with heterotopic bone formation in the elbow, after surgery.

Case report

A 9-year-old girl presented to the paediatric surgical emergency department approximately one hour after an injury to the right elbow. Typical X-ray imaging was performed and a diagnosis of displaced fracture of the lateral humeral condyle (Jacob, type II) was established. The fracture extended through the articular surface and there was moderate rotational displacement. After the standard pre-operative procedure, she underwent emergency operation. An anterior approach was utilised to avoid the posterior vascular pedicle to the fracture fragment. This is important to avoid avascular necrosis of the fragment. After open repositioning, fragment stabilisation was performed using 2 smooth Kirschner wires. Postoperatively, a plaster cast was used for two weeks at 90° of flexion and forearm supination. Three weeks after the injury, physical therapy was started. Despite physical therapy the patient had limited motility in her elbow. Four weeks after injury, X-ray examination showed stressed calcifications with an obscured boundary to the surrounding tissue (Fig. 1a). The flexion in her elbow was very restricted, and alkaline phosphatase value in serum was 796 U/L, the normal range in children 8-12 years old being 179-472 U/L. On X-ray examination, six months after the injury we can clearly see notable osteogenous formation like a bridge (Fig. 1b). The existence of heterotopic bone tissue was confirmed by computed tomography 3D reconstruction (Fig. 1c). On X-ray examination, eight months after the trauma, we could clearly see the radiographic appearance of a



Fig. 1

Heterotopic bone tissue formation in the elbow. a. X-ray image 4 weeks after trauma : stressed calcifications with obscured boundary to surrounding tissue ; b. X-ray image 6 months after trauma : osteogenous bridge-like formation ; c. Computed tomography 3D reconstruction : We could clearly see the radiographic appearance of a defined cortex ; d. Intra-operative finding : heterotopic bone tissue ; e. Control X-ray image after re-operation : no heterotopic bone tissue.

Discussion

Traumatic heterotopic ossification occurs in 10-20% of predisposed patients (3). Although there have been reports of spontaneous myositis ossificans, the process is initiated by trauma in 60 to 75% of cases (6). Most cases occur in the first three decades of life. The development of heterotopic ossification is independent of the patient's race or sex (4). The most common locations include the thigh, hip, upper arm, calf and foot (1, 6). The aetiology of traumatic heterotopic ossification remains uncertain. Migrated bone marrow cells have been suggested as a potential cause of osteogenesis in connective tissue (3-4). Other theories suggest muscle lesions or interstitial haemorrhagic foci as a potential cause of muscle degeneration, perivascular connective tissue proliferation, and subsequent bone metaplasia.

Heterotopic ossification may be due to an interaction between local factors and unknown systemic factors. The

defined cortex, and the alkaline phosphatase value in serum was 202 U/L. The girl underwent re-operation, when the heterotopic tissue was removed in its entirety (Fig. 1d). On the control X-ray a week after the surgery, is clearly evident that there is no longer any heterotopic bone tissue (Fig. 1e). Two weeks after the second operation the girl commenced physical therapy which lasted for six months. Two years after the injury the girl was re-evaluated : she had full motility of the elbow (Fig. 2a,b), and on control X-ray no heterotopic tissue was observed.

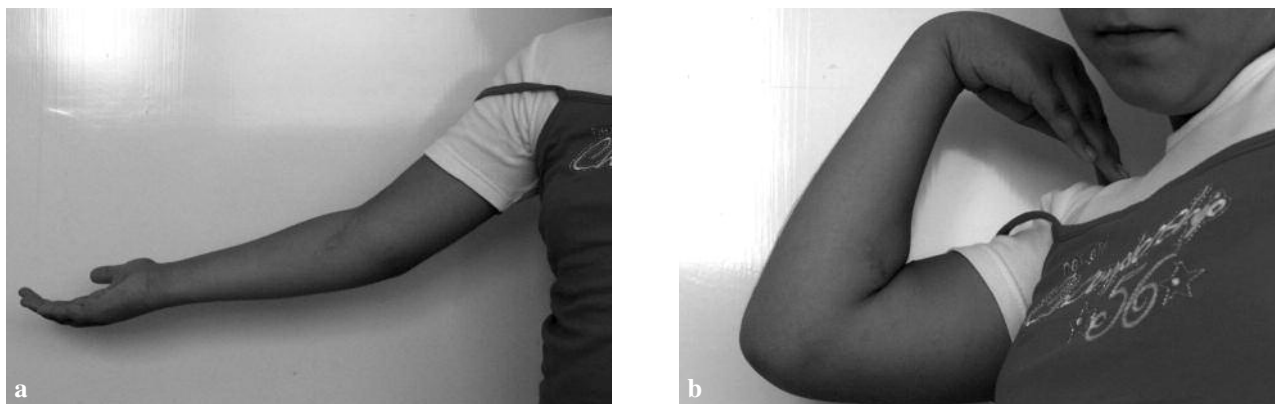


Fig. 2

Motility of the elbow two years after surgery. a. Extension ; b. Flexion

basic defect in heterotopic ossification is the inappropriate differentiation of fibroblasts into bone-forming cells.

However, various models exist, and it is thought that 3 conditions must be met for heterotopic ossification to develop : osteogenic precursor cells must be present ; an inductive stimulus should exist ; and the local tissue environment should be favourable.

Symptoms tend to be localised and usually consist of localised swelling, tenderness and decreased mobility of adjacent joints. The mature heterotopic ossification lesion often has a neocortex and medullar cavity (6), as was the case in our patient. Although it is rare, malignant transformation to osteosarcoma has been described (7).

Traumatic heterotopic ossification can be treated in a number of ways, but often it is quite challenging and, in many cases, with unsatisfactory results.

The role of physical therapy in patients with heterotopic ossification is controversial. The major goal of treatment is to maintain the range of motion and thereby preserve function. There are opposing philosophies about physical therapy. One theory is that an aggressive regimen of a passive range of motion exercises may predispose the patient to the development of heterotopic ossification because of micro trauma or local haemorrhage (2, 8). Several authors suggest that passive stretching and a range of motion exercises are contra-indicated after heterotopic ossification is suggested, but they recommend active exercise within the pain-free range (8, 9). Surgery for the removal of ectopic bone should be undertaken only for clear functional goals, such as improved standing posture or ambulation or for independent dressing and feeding. Excision should be considered for patients in whom shoulder motion is severely limited by extensive heterotopic bone ; heterotopic ossification that restricts elbow motion is excised surgically at maturation, as in our case. Maturation of heterotopic ossification is determined by the radiographic appearance of a

defined cortex and by a normal level of serum alkaline phosphatase. Some authors recommend excision after three months, although the bone is not matured. We prefer excision at maturation of the bone because the anatomy is much clearer. Prophylaxis against, or medical treatment of, heterotopic ossification is very controversial. This prophylaxis should be given in the form of non-steroidal anti-inflammatory agents, such as indometacin, during the early phase of heterotopic ossification and after excision (10). Other authors find that indometacin is not effective in preventing ectopic bone formation after surgery (11, 12). Radiation therapy may be effective if given 24 hours pre-operatively or within 72 hours postoperatively (10). In the literature there is no evidence about usage of indometacin or irradiation in the children population for prevention of heterotopic ossification.

Conclusion

Radical surgical excision of heterotopic ossification at maturation of the bone, following active physiotherapy around the elbow in children, can improve the arc of motion and the function of the extremity. Relatively simple operative and postoperative treatment can achieve satisfactory results.

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