CASE REPORT

Heterotrophic ossification causing Intraosseous ulnar nerve in elbow – A rare presentation in a rare problem!

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ABSTRACT

Background: Heterotrophic ossification, myositis ossificans, and periarticular calcification are different types of ectopic ossification.
Case Description: A 30-year-old male with recovered head injury and traumatic right brachial plexus injury presented with a reduction in range of movement of the right elbow due to ectopic ossification of the right elbow. He underwent surgical excision of the new bone and showed improved range of movement.
Conclusion: Heterotrophic ossification can cause deposition around nerves and encase the nerve, apart from causing movement restriction due to mechanical blockade. In spite of absent clinical signs of nerve involvement, the nerve might be entrapped in the ossification. Timely intervention with precise surgery under loupe magnification and tourniquet control prevents inadvertent nerve injury and gives good results.
Clinical Significance: The formation of new bone surrounding neurovascular structures must be recognised to prevent unintentional iatrogenic damage to the structures.
Keywords: Heterotrophic ossification, myositis ossificans, periarticular calcification, and ulnar nerve entrapment.
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INTRODUCTION

Heterotopic ossification (HO) is a process of benign bone formation and growth outside of normal skeletal locations. The prevalence of heterotopic ossification after central neurologic injury has been reported to range from 10% to 53%2. The elbow is the third most commonly affected joint following a traumatic brain injury[1]. When there is an ectopic ossification around the joint, it affects the range of movements and sometimes can present with symptoms of neurovascular compressions3. We report a patient who presented to us with heterotrophic ossification in the elbow following head injury and brachial plexus injury in the same limb.

CASE DESCRIPTION

30 A 30-year-old gentleman presented with difficulty in using his right elbow for 9 9-month duration. He met with a Road traffic accident one year ago and was diagnosed with a traumatic brain injury and right brachial plexus injury. He was hospitalised and managed conservatively. His workup revealed no fractures. He slowly recovered from the diffuse axonal injury to his brain. He was under regular follow-up as an outpatient, and his Brachial plexus injury recovered well within two months of the injury. He went back to his regular work. He presented to us one year after the initial injury with a gradual reduction in right elbow flexion. He did not have any comorbid conditions.

On examination, the patient's higher mental function was normal. His right elbow showed diffuse swelling in the medial aspect. The swelling was not mobile, not tender and bony hard in consistency. The elbow had a fixed flexion

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deformity of 30 degrees, and he was able to flex the elbow from 30 degrees to 90 degrees as shown in Figures 1a and 1b. The range of pronation-supination was full. The shoulder joint, wrist joint and joints of fingers and thumb had full range of pain-free movements. Both radial and ulnar artery pulsations were palpable. There was no neurological deficit of the ulnar, median and radial nerves. The sensations of the limb were intact.

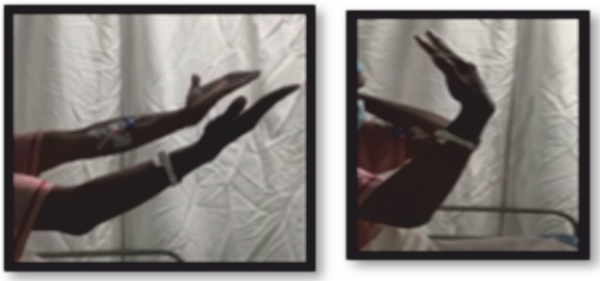


Fig. 1a & 1b: Pre-operative clinical image showing restriction of elbow movement.

The radiograph of the elbow revealed a bony shadow in the medial aspect of the elbow (Figure 2). He was planned for surgical excision. He underwent the procedure under general anesthesia and tourniquet control under loupe magnification. The operative findings are listed below-

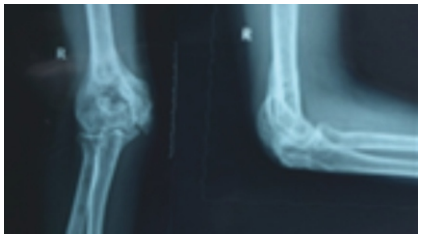


Fig. 2: Pre-operative x-ray of the elbow.

- The skin is incised longitudinally between the medial epicondyle and the olecranon, and sharp dissection of subcutaneous tissue is done with scissors.
- The roof of the cubital tunnel was found to be completely ossified, and the ulnar nerve was encapsulated by the new bone as shown in Figures 3a and 3b.



Fig. 3a &3b: Intra-operative image of the elbow showing the ulnar nerve surrounded by bone circumferentially.

- Excision of the osseous mass was done under C-arm guidance, and elbow arthrolysis was done.
- The new bone was soft and mostly nibbled, but the bone covering the ulnar nerve was hard and was removed carefully by chiselling.
- Flexion of range from 10 degrees to 140 degrees obtained on the table.
- The medial collateral ligament was repaired, and skin closure was done.

The patient was allowed to move his elbow on the second post-operative day. He didn't develop any ulnar nerve paresis. He developed seroma in the wound site, which was drained by needle aspiration and settled with conservative management. He underwent suture removal on the 14th post-operative day. The histopathological examination of the excised specimen revealed it to be bone and didn't provide any other information. He received regular therapy, and five months after surgery, he had a residual fixed flexion deformity of 10 degrees alone. He was able to flex actively from 10 degrees to 130 degrees (Figures 4a and 4b).



Fig. 4a & 4b: Post-operative clinical image showing improvement of elbow movement.

DISCUSSION

Ectopic ossification refers to the formation of pathologic bone and is an umbrella term encompassing heterotrophic ossification, myositis ossificans, and periarticular calcification. The heterotrophic and myositis ossification refer to the deposition of mature lamellar bone in nonosseous tissue and muscle, respectively. The periarticular calcification, though commonly grouped under heterotrophic ossification, refers to the deposition of calcium pyrophosphates in collateral ligaments and joint capsule4. Also radiographically, it does not display the trabecular organisation seen in both heterotrophic and myositis ossification. Our patient falls under the category of Periarticular calcification.

Heterotrophic Ossification can be conceptualised as

aberrant tissue repair and is increasingly recognised as a common complication of localised trauma, surgery, genetic, post-head and spinal cord injury and post thermal injury5.

Though multiple reports of ectopic ossification causing compression neuropathy of the brachial plexus and peripheral nerves are present in the literature, no case has been reported developing ectopic ossification in a limb that has recovered from traumatic brachial plexus injury. Though the pathophysiology of heterotrophic ossification is incompletely understood, it is believed to occur through inappropriate differentiation of fibroblasts into osteogenic cells[3]. Our clinical scenario shows that heterotrophic ossification can occur in a paralysed limb.

There are case reports of Ulnar nerve neuropathy7-11 due to myositis ossificans in the medial side of the elbow, but our patient didn't have any signs of ulnar nerve compression. In our patient, the medial collateral ligament ossification has led to the formation of new bone around the ulnar nerve. The common sites of ossification in the elbow are olecranon fossa, olecranon extension and humeroulnar bridge posteriorly, coronoid, humeroulnar, humeroradial and soft tissue anteriorly, medial collateral ligament medially and lateral collateral ligament laterally[4].

Though the normal range of flexion-extension of the elbow is 0°–145°, the functional range of motion required for daily activities is 30°–130° of flexion and extension and 50° of supination to 50° of pronation. Our patient had a loss of range of movement in flexion and extension axes but not in supination and pronation axes, falling under category of class IIA of Hastings and Graham's classification for Heterotrophic ossification at the elbow[2]. Most of the time, as the range of movement is reduced, we have to opt for surgical excision of the newly formed bone to improve the range of movement.

NSAIDs, Bisphosphonates and radiation can be used prophylactically in suspected patients[2]. Non-surgical methods like serial casting, static splinting, dynamic splinting, Continuous passive motion and manipulation are recommended in early stages alone[12]. Although the condition is said to be self-limiting, excision is unavoidable in circumstances where the myopathic mass limits daily activities[13]. We also had to resort to surgical excision in our patient. There are reports of the usage of Bisphosphonates after surgical excision to prevent new bone formation[14], but we didn't use any as the patient presented to us late.

The case is unique for developing ossification in the recovered brachial plexus palsy limb and for having bony hard ossification encompassing the ulnar nerve. It is notable that though the patient didn't have any pre-operative signs of ulnar nerve neuropathy, despite having an ulnar nerve surrounded by bone circumferentially.

CONCLUSION

Though heterotrophic ossification is a benign and self-limiting problem, it needs surgical excision of the new bone to restore the range of movements. Timely surgery and proper physiotherapy can provide good results.

Clinical Significance

Ectopic ossification of the elbow will need surgical intervention, and we have to be aware that the bone formation can be around neurovascular structures, especially the ulnar nerve. Precautions need to be taken during surgery to isolate and safeguard the neurovascular structures from injury.

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