

BRIDGING GLENOHUMERAL HETEROTOPIC OSSIFICATION AT THE KENYATTA NATIONAL HOSPITAL : A CASE REPORT

H.O. Ong'ang'o, MMed (Surg) , MSc Ortho (London), FCS-ECSA, Consultant Orthopaedic and Trauma Surgeon, Orthopaedics Department, Kenyatta National Hospital, and Honorary Lecturer, Department of Orthopaedic Surgery, University of Nairobi, Kenya and **F.W. Nang'ole**, MMed (Surg), Fellowship in Plastic and Reconstructive Surgery (Capetown), Fellow of the European Board of Plastic and Reconstructive Surgery, Lecturer in Plastic and Reconstructive Surgery, School of Medicine , University of Nairobi, Kenya

Correspondence to: Dr. H. O. Ong'ang'o, P.O. Box 74037-00200, Nairobi, Kenya. Email: herbertongango@yahoo.com

ABSTRACT

Heterotopic Ossification (HO) is defined as the process by which trabecular bone forms outside of the skeletal structure, occupying space in soft tissues where it does not normally exist. Three forms have been identified. There are traumatic, neurologic and the rare genetic forms of heterotopic ossification. It is a severe complication when it occurs. It can manifest with reduced range of joint movements, pain, stiffness and even compromised hygiene. It can also mimic malignant bone sarcomas. It is considered a very rare lesion in the shoulder. Management begins with prophylaxis in the individuals at risk by pharmacological means and/or irradiation. Surgery is reserved for those situations where the bone has matured and the benefits far outweigh the risks. Recurrence after surgery is a very common event. Prophylaxis after surgery for heterotopic ossification is highly recommended.

This case report is of a bridging posttraumatic glenohumeral heterotopic ossification of the left shoulder with a concomitant posterior axillary fold contracture which was treated by closed serial manipulations followed by contracture release, transpositional flap fashioning and split thickness skin grafting, showing good early postoperative functional results. There is no clear report to date of serial closed manipulation, transpositional flap fashioning and split thickness skin grafting in the literature on the management of heterotopic ossification of the shoulder.

INTRODUCTION

Heterotopic Ossification (HO) is defined as the process by which trabecular bone forms outside of the skeletal structure, occupying space in soft tissue where it does not normally exist (1-4). The aetiology continues to be elusive (1-3), but this is a known potential complication following trauma (4), blast injuries (4), elective surgery (5), neurological injury (6,7), severe burns (1,2,4,5,8,10,11), as well as some rare genetic conditions (3,4,5,11). In general, the most common sites for formation of heterotopic ossification is following Open-Reduction Internal Fixation (ORIF) of acetabular fracture (1,2), followed by the hip after total hip arthroplasty (1,2), the elbow after distal humerus fractures and fracture-dislocations (4,11) and at the knee after total knee arthroplasty (1). At the shoulder, heterotopic ossification is considered rare (6,7,13-15). Nevertheless heterotopic ossification at the shoulder has been reported after encephalitis (16), bilateral intradeltoid injections (17), bilateral hemiarthroplasty (19), cuff tear arthropathy (19), hemiarthroplasty (13), total shoulder arthroplasty (13,14,18), reverse total

shoulder arthroplasty (14), arthroscopic acromioplasty and rotator cuff repairs (10,15) as well as after distal clavicle resections (10). Shoulder heterotopic ossification has also been reported after chronic sepsis and coma (15) and also as a complication of all forms of treatment for displaced three-part and four-part fractures of the proximal humerus (20).

Clinically, heterotopic ossification will manifest with severe reduced range of joint movements (8-11,13,17), pain (11,13), stiffness (11,17) and sometimes compromised hygiene (15).

Traditionally, management commences with prevention in those predisposed by pharmacological agents such as NSAIDs (1) and bisphosphonates (8) as well as prophylactic irradiation (5, 15). NSAIDs that have been used in the prophylactic treatment of heterotopic ossification include indomethacin, naproxen, celecoxib, rofecoxib, etoricoxib, diclofenac, ibuprofen, aspirin, ketoprofen, and piroxicam (10). The gold standard in NSAID therapy of heterotopic ossification remains indomethacin (1). Bisphosphonates use mainly centered around etidronate but now they are no longer used in prevention of heterotopic

ossification (2,5). Newer pharmacological modes of prevention and treatment that give fewer side effects than indomethacin and radiation therapy are on trial. These include noggin, didronel, apyrase, free radical scavengers (Allopurinol and N-Acetylcysteine) (1). Exposure to Pulsed Electromagnetic Fields (PEMF) is another new method on trial for the prevention and treatment of heterotopic ossification (1).

Surgery is advocated for treatment of established heterotopic ossification (9). Surgery is advised for the established heterotopic ossification where there is interference with movements, pain, nerve entrapment and situations of compromised hygiene (4,11). Surgery is further acknowledged as the only treatment that is capable of removing already formed bone (2) and it can be carried out in a closed, minimally invasive, arthroscopic or open fashion (10). Recurrence of heterotopic ossification after surgery is a well documented event (5). A caveat has been put to emphasize that surgery should be undertaken only if the expected benefits clearly outweigh the risks (3).

We present a case report of a patient who had bridging glenohumeral heterotopic ossification severely restricting left shoulder movements and whose management involved repeated closed bone lysis followed by contracture release, transpositional flap fashioning and split thickness skin grafting with good early postoperative functional results.

CASE REPORT

Sometimes in the middle of the year 2012, a 29 year old African male presented to us at The Kenyatta National Hospital with history of left shoulder injury of one year duration. He was experiencing limited left shoulder movements. No further details on the nature of the shoulder injuries sustained nor the treatment received elsewhere were forthcoming. Examination revealed a young man in very good general condition. The shoulder examination revealed wasted shoulder muscles, a prominent lateral end of the left clavicle and a 3 millimetres mobility of the left acromioclavicular joint (Figure 1). There was also a scar in the axilla involving the posterior axillary fold. Testing for shoulder movements showed that he had active elevation of 20 degrees in the left shoulder, while active external rotation was 0 (ZERO) degrees (Figure 2).

Review of the X-rays showed the patient had; (A) heterotopic ossification bridging the glenoid and surgical neck of humerus; (B) heterotopic ossification outer 1/3rd of left clavicle and (C) a subluxated left

acromioclavicular joint (Figures 3 and 4). X-rays of the contralateral right shoulder showed a perfect shoulder joint (Figure 5).

At this point a diagnosis of bridging left glenohumeral heterotopic ossification, corresponding to Grade III (13,14,16) was made and patient was commenced on pharmacological treatment. This comprised of capsules of indomethacin 25 mg three times daily and physiotherapy. A plastic and reconstructive surgeon was consulted and preoperative workup was done. Within a week, the patient underwent the first episode of closed manipulation in theatre under general anaesthesia, when closed bone lysis was achieved and an active elevation of 80 degrees recorded postoperatively. Post manipulation radiographs revealed fracture of the heterotopic ossification bone mass (Figure 6). When pain had subsided, the patient received physiotherapy which was reported as maintaining the achieved elevation. After an interval of 19 days the patient underwent a second episode of closed manipulation under general anaesthesia and this extended the achieved active elevation to 100 degrees (Figure 7). Patient again resumed physiotherapy when the pain had subsided. Two weeks after the second manipulation, the patient underwent plastic and reconstructive surgical intervention. This consisted of release of contracture, transpositional flap fashioning and split thickness skin grafting. After a rest period the patient resumed physiotherapy. Shortly after this he had attained 120 degrees of active elevation and he sought and was granted discharge from our facility. All along he was on oral indomethacin 25 mg three times daily. He was discharged on oral indomethacin 25 mg three times daily. Sadly, the patient has not been seen again.

Figure 1

Wasted musculature of the left shoulder



Figure 2

Left shoulder with only 20 degrees of elevation



Figure 5

An axillary view X-ray of the contralateral normal right shoulder



Figure 3

AP view of X-ray of left shoulder showing bridging bone from surgical neck of humerus to the glenoid



Figure 6

Post operative X-ray. The heterotopic ossification bone mass has been fractured and 80 degrees of elevation achieved after initial closed manipulation

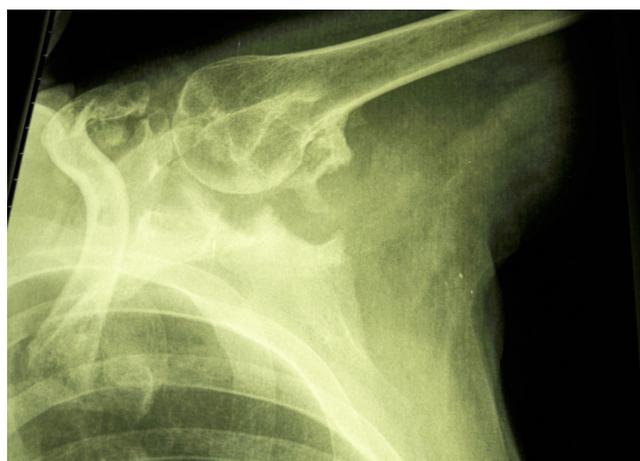


Figure 4

Axillary view X-ray of the left shoulder again showing the bridging bone



Figure 7

100 degrees of elevation after two closed manipulations



DISCUSSION

That heterotopic ossification is a serious complication is well acknowledged (2). Also acknowledged is the rarity of this condition around the shoulder (6, 11,13,15,19) and the fact that it has been rarely studied (7,9). In our patient, heterotopic ossification followed trauma to the shoulder region. With the details of the exact nature of trauma not forthcoming and the exact duration since the trauma lacking, we were disadvantaged in making the diagnosis. All these factors added up to make this case a serious diagnostic challenge. We nevertheless did postulate that the primary injury was treated by immobilization and later forcible mobilization which are known triggers of heterotopic bone formation (2,10,11).

The patient presented with extreme limitation of shoulder movement. In assessing the shoulder movements at diagnosis we elicited positive findings in elevation and external rotation. This is not unique. Kjaersgaard-Andersen *et al* (16) also in their series mainly evaluated elevation with minor attention to internal and external rotation. With elevation of 20 degrees and external rotation of 0 degrees, severe limitation of shoulder movements was well illustrated (Figure 2). The 20 degrees of elevation was solely from scapulothoracic motion. Our patient also presented with a soft tissue contracture involving the posterior axillary fold. This also contributed to the limitation of joint movement. Existence of concomitant soft tissue contracture and heterotopic ossification has been described before (8). In summary, the severe disability shown by this case is itself a rarity similar to what Pansard *et al* (6) and Kjaersgaard-Andersen *et al* (16) have observed.

The only diagnostic imaging investigations performed on our patient were plain radiographs (Figures 3 - 7). The plain radiographic findings were unequivocal. The finding of mature bridging glenohumeral heterotopic ossification fitting in Kjaersgaard-Andersen *et al* Grade III (16) obviated the need for other investigations employed in heterotopic ossification. Thus ultrasound, 99technetium bone scans, computed tomographic scans, magnetic resonance imaging as well as single photon emission CT (SPECT) were deemed unnecessary. This partly resembles the series of Pansard *et al* (6) and that of Fuller *et al* (7) where bone scans were not used. Even alkaline phosphatase determination (3) was considered not necessary in this patient. The patient was commenced on indomethacin promptly as arrangements to take him to theatre were being made. This was mainly as prophylaxis for any heterotopic

ossification that may recur after surgical intervention. We considered it prudent to begin with closed manipulation and then assess the results. The results of one episode of closed manipulation were very good. 80 degrees of active elevation were achieved and this encouraged us to perform a second episode of closed manipulation. Closed manipulation with success in shoulder heterotopic ossification has been described (15). What has not been described before is serial closed manipulations yielding success in heterotopic ossification of the shoulder. After evaluating the results of our two closed manipulations of the shoulder, we concluded that there was no need for open resection in that shoulder. We thus avoided open resection and its attendant complications (3). At this point we handed over the patient to our plastic and reconstructive surgery counterpart who performed release of contracture, fashioning of a transposition flap and split thickness skin grafting.

The timing of contracture release to come after surgical attention to the heterotopic bone was most appropriate as contractures have been observed to be difficult to treat preoperatively (6). After a rest period to allow for take of these procedures, the patient resumed physiotherapy. Final assessment showed he had attained elevation of 120 degrees. This gain in motion and function was very commendable given that in heterotopic ossification even small gains in motion and function are significant (7). No intra-operative iatrogenic humeral fracture was sustained while operating on this late heterotopic ossification with ankylosis (8).

All this time, the patient was maintained on indomethacin 25 mg three times daily. Right from the outset, radiotherapy was not considered as we contemplated the plastic and reconstructive procedures. We wished to avoid the wound healing delays associated with radiotherapy (4). The patient was finally discharged on indomethacin 25 mg three times daily and physiotherapy.

REFERENCES

1. Baird, E. O. and Kang, Q. K. Prophylaxis of heterotopic ossification. *J Orthop. Surg Res.* 2009; **4**(12): 1-8.
2. Bossche, L.V. and Vanderstraeten, G. Heterotopic ossification: A review. *J Rehab Med.* 2005; **37**: 129-136.
3. Shehab, D., Elgazzar, A. H. and Collier, B. D. Heterotopic ossification. *J Nuclear Med.* 2002; **43**(2): 346-353.
4. Nauth, A., Giles, E., Potter, B. K., *et al.* Heterotopic ossification in orthopaedic trauma. *J Orthop Trauma.* 2012; **26**(12): 684-688.

5. Balboni, T.A., Gobezie, R. and Mamon, H.J. Heterotopic ossification: Pathophysiology, clinical features, and the role of radiotherapy for prophylaxis. *Intern J Radiation Oncol Biol Physics*. 2006; **65**(5): 1289-1299.
6. Pansard, E., Schnitzler, A., Lautridou, C., *et al*. Heterotopic ossification of the shoulder after central nervous system lesion: indications for surgery and results. *J Shoulder Elbow Surg*. 2013; **22**: 767-774.
7. Fuller, D.A., Mani, U.S. and Keenan, M.A. Heterotopic ossification of the shoulder in patients with traumatic brain injury. *J Shoulder Elbow Surg*. 2013; **22**: 52-56.
8. Hsu, J.E. and Keenan, M.A. Current review of heterotopic ossification. *University of Pennsylvania Orthop J*. 2010; **20**: 126-130.
9. Boynton, M.D. and Enders, T.J. Severe heterotopic ossification after arthroscopic acromioplasty: A case report. *J Shoulder Elbow Surg*. 1999; **8**: 495-497.
10. Berg, E.E. and Ciullo, J.V. Heterotopic ossification after acromioplasty and distal clavicle resection. *J Shoulder Elbow Surg*. 1995; **4**: 188-193.
11. Kaplan, F.S., Glaser, D.L., Hebela, N., *et al*. Heterotopic ossification. *J Amer Academy Orthop Surg*. 2004; **12**: 116-125.
12. Guitton, T.G., Duckworth, A.D., McQueen, M.M., *et al*. Elbow dislocation and articular fracture of the distal humerus. *Shoulder and Elbow*. 2010; **2**: 111-117.
13. Boehm, T.D., Wallace, W.A. and Neumann L. Heterotopic ossification after primary shoulder arthroplasty. *J Shoulder Elbow Surg*. 2005; **14**(1): 6-10.
14. Wierks, C., Skolasky, R.L., Ji, J.H., *et al*. Reverse total shoulder replacement. *Clin Orthop Relat Res*. 2009; **467**: 225-234.
15. Warner, J.J.P. Ejnisman, B. and Akpinar, S. Surgical management of heterotopic ossification of the Shoulder. *J Shoulder Elbow Surg*. 1999; **8**: 175-178.
16. Kjaersgaard-Andersen, P., Frich, L.K., Sojbjerg, J.O., *et al*. Heterotopic bone formation following total shoulder arthroplasty. *J Arthroplasty*. 1989; **4**: 99-104.
17. Mylle, J., McCombe, P. and Weatherley, C. Bilateral intradeltoid ossification: A cause of glenohumeral abduction contracture. *Acta Orthop Belgica*. 1991; **57**(2): 195-198.
18. Sperling, J.W., Cofield, R.H. and Rowland, C.M. Heterotopic ossification after total shoulder arthroplasty. *J Arthroplasty*. 2000; **15**(2): 179-182.
19. Cheah, J., Nigro, P., Smith, E., *et al*. Shoulder heterotopic ossification after bilateral hemiarthroplasty: case report and review of the literature. *J Shoulder Elbow Surg*. 2011; **20**: e7-e13.
20. Neer, C.S. II. Fractures of the proximal humerus. Part II. Treatment of three-part and four part displacements. *J Bone Joint Surg. Am*. 1970; **52A**(6): 1090-1103.