

Case Report

DOUBLE JEOPARDY : PATHOLOGICAL FRACTURE AND NON-UNION OF PROXIMAL HUMERUS DUE TO CHONDROCALCINOSIS

Vikram Shetty¹, Siddharth M Shetty², Arjun Ballal³, Mayur M Kamani⁴ & B Jayaprakash Shetty⁵

^{1,2}Associate Professors, ^{3,4}Postgraduate, ⁵Professor, Department of Orthopaedic Surgery, K.S. Hegde Medical Academy, Nitte University, Deralakatte, Mangalore, Karnataka, India.

Correspondence :

Arjun Ballal

Postgraduate, Department of Orthopaedic Surgery, K.S. Hegde Medical Academy, Nitte University, Deralakatte, Mangalore, Karnataka, India.

Mobile : +91 84316 44077 E-mail : 5arjunballal@gmail.com

Abstract :

Fractures of the humerus constitute 5% to 8% of fractures and most have an uneventful healing, but occasionally non-unions of the fracture with joint stiffness of shoulder and/or elbow and prolonged debilitating pain may be encountered. Predisposing conditions usually are osteoporosis, obesity, alcoholism and smoking. Comminuted or segmental fractures, soft tissue interposition at the fracture site, improper fixation and infection may also result in non-union.

A fracture of neck of the right humerus (dominant limb) in a young lady failed to unite following internal fixation and lead to atrophic non-union with implant loosening, which initially was suspected to be due to poor fixation and or infection, but histopathological evaluation on two separate occasions of the fracture site revealed a picture of Chondrocalcinosis (CC). CC is a condition wherein deposition of calcium pyrophosphate dihydrate (CPPD) crystals occurs within articular cartilage and synovial tissue and is associated with joint pain leading to arthritis. The presence of these CPPD crystal has not been mentioned in the bone or at fracture sites in the literature, The histopathological evidence of CPPD at the fracture site in this case of humerus non-union did not establish whether the fracture was due to the pathology or the CPPD got deposited at the fracture site and was a cause of non-union.

Keywords : Chondrocalcinosis, Calcium pyrophosphate dehydrate(CPPD) ANKH gene, PHILOS, Non union.

Introduction:

We report this case of Calcium Pyrophosphate Dihydrate(CPPD) deposition at fracture site of humerus which progressed to non-union. Occurrence of CPPD deposition in bone is very rare. We have failed to conclude whether the CPPD deposition at the fracture site was a cause of non-union or was primarily a cause of pathological fracture. Introduction: Chondrocalcinosis is a Calcium pyrophosphate dihydrate (CPPD) crystal deposition disorder primarily involving the articular cartilage and the synovial tissue in and around the joint¹. In our case we

observed the deposition of crystal at the fracture non-union site of the humerus neck. This occurrence of chondrocalcinosis at an extra articular site like in metaphyseal area of a

bone and in a non-union site of a fracture is a rare occurrence with only one case being reported in the literature⁵.

Case History :

A 35 year old lady presented to us with pain and instability of the dominant shoulder for eight months. She had sustained an injury following a fall eight months ago and was diagnosed with a fracture right proximal humerus(Fig:1). She underwent open reduction internal fixation with PHILOS plate (Fig:2) at a different hospital and was on regular follow up with the primary treating surgeon. She reported to us after eight months of surgery with pain and instability of the right shoulder. She had tenderness and abnormal mobility of the proximal third of humerus with restricted active and passive range of motion of the right shoulder with pain and crepitus. The surgical wound had healed with primary intention. Radiographs revealed a fracture non-union of right humerus neck and loosening of PHILOS implant with calcific specs around the fracture

Access this article online

Quick Response Code





Figure-1 : Fracture neck of humerus day one



Figure-2 First post op



Figure-3 : Loosening of implant noted at 6 months post op

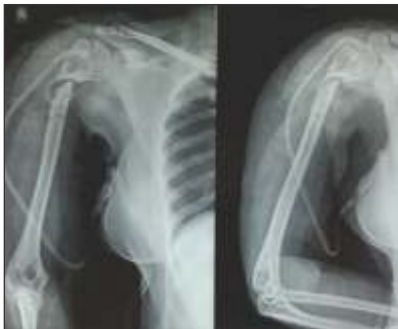


Figure - 4 : After removal of implant -post op



Figure-5 : Six months after after removal of implant, no signs of union noted



Figure-6 : Immediate post op, after PHILOS fixation and bone grafting (reosteosynthesis)



Figure-7 : Follow up after 10 months. Signs of healing noted with minimal callus formation

site(Fig:3) and haematological reports ruled out infection. She was planned for implant removal, debridement, bone grafting and re-osteosynthesis. The patient was operated under General Anaesthesia through the previous incision and implant was removed, and chalky white deposits were noted at the fracture site with no evidence of infection. The tissue from the fracture non-union site was sent for analysis and in view of such tissue being observed at fracture site re-osteosynthesis was not performed (Fig:4). The biopsy reports revealed Chondrocalcinosis. The case was discussed with the primary treating surgeon regarding

use of synthetic bone graft substitutes during primary procedure, which was denied by him, as in this case. She was immobilized in an arm pouch for 6 months. At 6 months the X ray revealed non-union at the fracture site with calcific speckling in the soft tissues adjoining the non-union site (Fig:5) and routine blood investigations were normal. She was operated upon the non-union site was freshened, the tissue was sent for histopathological examination, with autologous corticocancellous bone grafting and internal fixation with PHILOS plate(Fig:6). The histopathology was reported as chondrocalcinosis. She was on regular follow up, at six months there was delayed progression in fracture healing, and at ten months from the second surgery the range of motion of shoulder improved and as per Constant Murley score it had improved from score of seven at time of first visit to 33 with no pain or instability, the radiographs shows the implant is in good position, the fracture healing has progressed but not complete and calcific speckling has resolved partially (Fig:7).

Discussion :

Chondrocalcinosis is a disorder of calcium pyrophosphate crystal deposition (CPPD) in the articular cartilage, synovial tissues lining the joint and the tendon sheaths adjacent to the joints.¹ Its occurrence has not been reported in osseous structures. Chondrocalcinosis can occur in a Sporadic form or as in secondary chondrocalcinosis. Sporadic is a common occurrence in the elderly and often associated with osteoarthritis² and also with hereditary haemochromatosis, hyperparathyroidism and

hypomagnesemia are metabolic disorders that predispose to secondary chondrocalcinosis.³ This usually is attributed to a genetic predisposition to such condition which is a mutation in the ANKH gene that is involved in the transport of inorganic pyrophosphate.¹ Fang HQ and colleagues reported an intra-articular chondrocalcinosis in the knees of 20 patients⁴. Agrawal and colleagues reported a case of extra-articular chondrocalcinosis in a 53 year old man in the right scapular area which was initially considered to be a sarcoma.⁵

References :

1. Adrian Pendleton, Michelle D. Johnson, Anne Hughes, Kyle A. Gurley, Andrew M. Ho, Michael Doherty; Mutations in ANKH cause chondrocalcinosis. *Am J Hum Genet.* Oct 2002; 71(4): 933–940.
2. Abhishek A, Doherty S, Maciewicz R, Muir K, Zhang W, Doherty M. "Evidence of a systemic predisposition to chondrocalcinosis and association between chondrocalcinosis and osteoarthritis at distant joints: a cross-sectional study". *Arthritis Care Res (Hoboken).* 2013 Jul;65(7):1052-8.
3. Richette, P., Bardin, T. & Doherty, M. An update on the epidemiology of calcium pyrophosphate dihydrate crystal deposition disease. *Rheumatology (Oxford)* 48, 711–715 (2009)
4. Fang HQ, Li QM, Huang YQ, Xing JS, Mao RJ, Xie L. "Calcium pyrophosphate dihydrate crystal deposition disease: a clinicopathologic analysis of 20 cases". *Zhonghua Bing Li Xue Za Zhi.* 2012 Dec;41(12):828-32.
5. Agrawal A, Purandare N, Sridhar E, Shah S, Dua SG, Rangarajan V. "Imaging findings in a rare case of extra-articular chondrocalcinosis". *Clin Nucl Med.* 2012 Feb;37(2):184-7.