# **Global Hypertrophic Calcification of Shoulder Joint Capsule**

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# ABSTRACT

BACKGROUND: Calcification around the shoulder joint usually occur inside or around the tendons of the rotator cuff. We herein report on a case of global hypertrophic calcification of shoulder joint capsule in a patient with Rheumatoid arthritis.

CASE REPORT: An 86 years-old male with a long-standing history of seropositive Rheumatoid arthritis. The treatment for his Rheumatoid arthritis included Methotrexate and Hydroxychloroquine initially, but due lack of control, adalimumab was added with excellent control of his arthritis. He has progressively experienced an increasing pain and stiffness in his shoulders, in addition to an increasing limitation of shoulder movement. Magnetic Resonance Imaging revealed severe arthritis with remoulding deformity with extensive capsular calcification, intraarticular loose-bodies.

DISCUSSION: This phenomenon of calcification of shoulder capsule has not been reported before. The pathophysiology of calcific tendinopathy of the shoulder remains controversial. The calcific deposits consist of poorly-crystallized hydroxyapatite.

**CONCLUSION:** Global hypertrophic calcification of shoulder joint capsule is unique and unreported in the literature. We can postulate that the long-standing inflammation of the synovial lining of the capsules had a major part. Moreover, Diabetes Mellitus, smoking, and repetitive manoeuvres are recognized contributing factors as well for similar conditions. Genetic predisposition seems to play a role as well. We think all those have played part in the development of this unprecedented presentation. Management should be tailored to target specific symptoms for pain, rigidity, and decreasing calcification size. Several options are available, including Kinesiotherapy, electrotherapy modalities, iontophoresis, electroshock wave therapy, and finally surgical approaches for progressive and refractory cases.

KEYWORDS: Rheumatoid arthritis, calcific tendinitis, Hydroxyapatite, crystal

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# Background

Calcification around the shoulder joint usually occur inside or around the tendons of the rotator cuff; more specifically, within the supraspinatus, infraspinatus tendons and subacromial burse.<sup>1,2</sup> Its prevalence is estimated to be around 2.7% to 10.3%.<sup>3-6</sup> Unlike those reported before, which usually affects the tendons around the joint, we herein report on a case of global hypertrophic calcification of shoulder joint capsule in a patient with rheumatoid arthritis. This manifestation has not been reported before.

# **Case Report**

An 86-year-old male with a history of seropositive rheumatoid arthritis for 30 years. The treatment for his rheumatoid arthritis included methotrexate initially, but due lack of control, adalimumab 40 mg subcutaneously every 2 weeks was added with excellent control of his arthritis of hands and feet. This lasted for few years. However, he later on experienced activity of disease and he was switched from adalimumab to Tocilizumab 162 mg subcutaneous injections every 1 week. His arthritis was controlled on the latter regimen apart from his shoulders. At this time his lab investigations were normal including complete blood panel (CBC), C-reactive protein (CRP), erythrocyte

sedimentation rate (ESR), Liver function test (LFT), Renal function test (RFT), and Bone profiles. He experienced an increasing pain and stiffness in his shoulders, in addition to an increasing limitation of shoulder movement in all directions. His other medical conditions include type 2 diabetes mellitus and hypertension. We are unaware of any history of adhesive capsulitis. His other medications are Methotrexate 10 mg oral once weekly, hydroxychloroquine 200 mg, alendronate 70 mg once weekly. X-rays of shoulders at the time (May 2012 and are shown in Figure 1). They showed punctate and linear calcification around the capsule. The patient received multiple intraarticular steroid injections with no notable benefit. Over the years of follow-up visits, his condition progressed, with increasing limitation of his shoulders movements. The Oxford Shoulder Score (OSS) were used to assess degree of pain and disability. Score for both shoulders were 7, indicating a great degree of disability.<sup>7</sup> Repeated imaging on March 2017 was performed and is shown in Figure 2 and showed hypertrophic, global plaque-like calcification in the capsule. Meanwhile, X-ray of hands did not reveal any chondrocalcinosis or erosions and knee x-rays showed advanced osteoarthritis. Magnetic Resonance Imaging (MRI) of left shoulder was done in November 2020 and is shown in Figure 3 and revealed severe

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Figure 1. Punctate and linear calcification around the capsule at baseline.



Figure 2. Calcific deposits in both shoulders at 5 years follow-up.

arthritis with remoulding deformity with extensive capsular calcification, intra-articular loose bodies and large joint effusion resulting in dislocation. In addition, he had severe rotator



Figure 3. Revealed severe arthritis with remoulding deformity with extensive capsular calcification, intra-articular loose bodies and large joint effusion resulting in dislocation.

cuff which atrophy reflects chronic complete tear with retraction and atrophy. The patient was offered surgical intervention, but he refused.

#### Discussion

In this report, we described a long-standing case of seropositive rheumatoid arthritis who gradually developed a global hypertrophic calcification of shoulder joint capsules. This phenomenon of calcification of shoulder has not been reported before. We can only postulate that the long-standing inflammation of the synovial lining of the capsules had a major part in its development. The bigger issue of what causes calcification around the joints in general is still controversial. The usual sites of calcification around the joint are the tendons and ligaments. The prevalence of the calcific tendinopathy has been reported to be 3% to 10% in the general population,<sup>3-6</sup> and 7% to 17% among individuals with shoulder pain.<sup>8</sup> We could not find any reports on calcification of joint capsules.

The pathophysiology of calcific tendinopathy of the shoulder remains controversial. The calcific deposits consist of poorly crystallized hydroxyapatite. Earlier on it was proposed that calcification is a consequence of age-related tendon degeneration, but this is not supported by several findings, including the relatively young patient age at the peak incidence, the spontaneous healing and resolution that occurs in many cases and the development of the condition in healthy, untraumatized tissue. Also, tendons overuse has been recognized by many authorities as another theory for calcification of the shoulders.<sup>9</sup>

In 1997, Uhthoff and Loehr<sup>10</sup> proposed that calcific tendinopathy was a disease that progressed through 4 distinct phases, with correlating pathologic and clinical findings. It typically starts with pre-calcific stage, which is usually asymptomatic, followed by calcific-stage, which can furtherly divide into formative and resorptive phases. Finally, post-calcific stage, in which collagen remodelling and tendon repair take place.<sup>10,11</sup> Some reports suggest that there is a genetic predisposition for the development of intratendinous calcific deposits, a claim supported by the relatively high frequency of bilateral occurrence. However, no clear proof of this has been advanced. Moreover, Diabetes Mellitus, smoking, and repetitive manoeuvres are recognized contributing factors as well.<sup>11</sup> In our case the patient was diabetic elderly gentleman with a long standing seropositive rheumatoid arthritis. We think all those have played part in the development of this unusual and hitherto unreported presentation.

While such burden of calcification is rarely encountered, the management expertise is largely deficient and unaccustomed. Beside conservative rehabilitation, several modalities have been implied with main guidance of its use depends on the following specific goals: pain, rigidity, and decreasing the size of calcification. Generally, a combination of these measures is usually employed. For pain, physical therapy, NSAIDs (non-steroidal anti-inflammatory drugs) are initially measures, before using electrotherapy with its diverse modalities such as microwaves, short waves, TENS (transcutaneous electrical nerve stimulation), ultrasounds, Interferential, pulsed electromagnetic. For rigidity, Kinesiotherapy, has proven to be useful and considered first option of treatment.<sup>12</sup> For management of the calcification, there are many options, such as iontophoresis, ESWT (electroshock wave therapy), ultrasound guided needle aspiration. Finally, surgical approach has been reserved for patients with progressive and refractory symptoms, particularly when all conservative treatments fail. Arthroscopic approach is currently gaining preference over open surgical technique.<sup>11,12</sup>

# Conclusion

Global hypertrophic calcification of shoulder joint capsule is unique and unreported in the literature. While contributing factors are not clearly recognized, long standing history of inflammatory arthritic condition and genetic predisposition seem to have a main role. Also, concomitant disorder such as Diabetes Meletus might be contributing. We think all those have played part in the development of this unprecedented presentation

#### **Patient Consent**

Was secured to publish the findings of this case.

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