Clinical, MRI And Histological Appearances Of Multiple Rice Bodies Formation In The Subacromial-Subdeltoid Bursa In Association With Polymyositis – Case Report And Literature Review

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Citation

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Abstract

We illustrate one case of spontaneous multiple rice bodies formation in a middle-aged lady with history of polymyositis, presented as painless cystic shoulder swelling, which is, to our knowledge, the first reported case in literature studies.

INTRODUCTION

The purpose of this article is to illustrate a case of spontaneous multiple rice bodies formation in a patient with polymyositis. Despite uncertain aetiology, rice bodies have been reported to be associated with tuberculosis and rheumatoid arthritis1. The condition is rare and only case reports exist in literature studies. Operative synovectomy or bursectomy is the mainstay of treatment, with excellent clinical outcomes and minimal recurrence.

CASE REPORT

A 60-year-old-woman was referred from Rheumatologist for orthopaedic consultation of left shoulder swelling. Background history included polymyositis manifesting as fever and proximal muscle weakness, diagnosed two years ago by muscle biopsy and electrophysiological studies. She was initially put on oral steroid, then switched to mycophenolate mofetil six months later for adequate control of elevated muscle enzymes and interstitial lung disease. Disease control was satisfactory with little clinical serological deterioration.

She reported insidious onset of asymptomatic left shoulder swelling for three months, and denied history of injury, injection, acupuncture or operation. C-reactive protein was 20mg/L, ESR was 60mm/hr, and other autoimmune markers including rheumatoid factor, anti-CCP and ANA were negative. Bedside aspiration was performed thrice by physicians, yielding clear straw-coloured fluid. All samples were negative for bacterial, fungal, and acid-fast bacilli cultures, and microscopy only showed a small number of inflammatory cells. The cyst quickly recurred in size after aspiration.

Physical examination showed a 12cm anterior and 7cm posterior shoulder cystic swelling, both being soft, smooth, non-tender and non-pulsatile. Both cysts became less apparent upon tensing up the deltoid and pectoralis muscle. (Fig. 1) The cysts were mobile in relation to skin and muscle, and could be emptied from one to another. Trans-illumination test was negative. Functional assessment of shoulder joint including motion range, power, and impingement signs were normal.

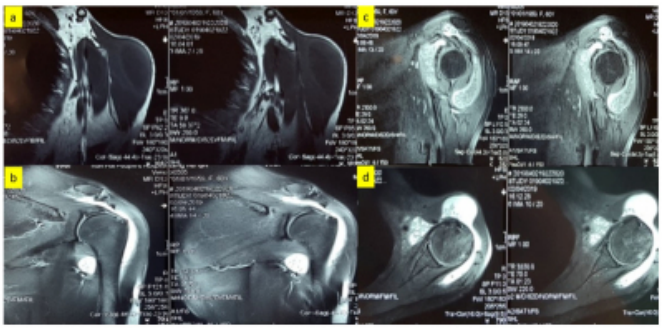
Figure 1

Clinical photo of left anterior shoulder swelling: The cyst became less apparent upon tensing up the deltoid and pectoralis muscle, indicating its submuscular location.

Non-contrast magnetic resonance imaging (MRI) of left shoulder showed a large fluid collection with multiple filling defects in the subacromial, subdeltoid and subcoracoid bursae. No septum was seen. The filling defects ranged 2 to 3mm in size, and were T1 isointense and T2 hypointense in nature. Rotator cuff and glenohumeral joint appeared normal. (Fig. 2)

Figure 2

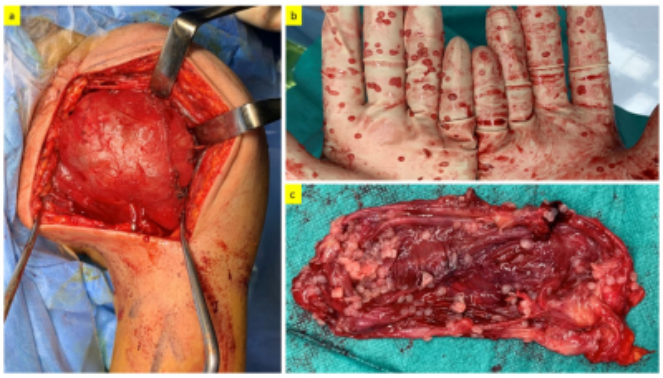
MRI imaging of the left shoulder:Left shoulder imaging showed large cystic fluid collection over the subcoracoid, subacromial and subdeltoid bursa, with numerous small filling defects. The filling defects were isointense on T1 and hypointense on T2 imaging. a. Coronal T1 image showing isointense rice bodies and shoulder bursa. b. Coronal T2 image showing intact supraspinatus muscle and tendon. c. Sagittal T2 image showing abundant rice bodies in the bursae. d. Axial T2 image showing bursal inter-connection and sparing of glenohumeral joint.



Open bursectomy was performed via deltopectoral approach. A large subacromial bursa with encapsulated fluid overlying the anterior glenohumeral joint was excised. No feeding stalk, vessels or nerve connections were noted. There was no communication with the glenohumeral joint or rotator cuff muscles. The outer surface was smooth, whilst the inner lining was thickened and folded. Hundreds of 2 to 3mm glistening granules, roundish smooth and firm in consistency, were found inside the cyst. The majority of the granules were free-floating, while some were attached to the synovial lining of the bursal sac. (Fig. 3)

Figure 3

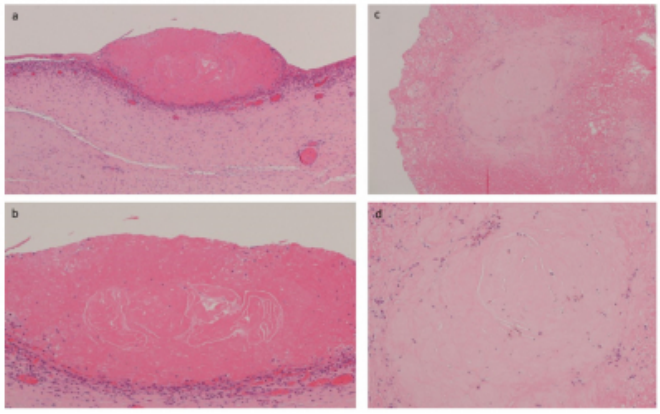
Intra-operative findings of the enlarged bursa and rice bodies. Hundreds of rice bodies were found inside the bursa, majority being free-floating while some attached to the inner lining. a. The presence of bursal cyst located over anterior shoulder. b. Appearance of free-floating rice bodies. Multiple glistening granules around few millimeters in size were drained. c. Appearance of inner cyst contents. Some rice bodies were seen attached to the inner cyst wall, which was reddish, folded and mild inflamed in appearance.



Pathology of the cyst wall showed focal synovial hyperplasia, increased capillary proliferation and mild mononuclear cells infiltration, consistent with bursitis. Section of the rice bodies showed central pauci-cellular eosinophilic materials surround by fibrin, with no chondrocytes nor osteochondral components. Congo red stain for amyloid was negative. Extended culture of the cyst wall and contents were negative for bacteria, fungus or acid fast bacilli. (Fig. 4)

Figure 4

Pathological sections of the bursal wall and the rice bodies. a. The shoulder cyst contains fibrotic cyst wall lined by synovium, with increased capillary proliferation and mild mononuclear cells infiltration. A small rice body is noted sitting on the luminal side of the cyst wall. b. The rice body on the luminal surface of fibrotic cyst wall is composed of mostly fibrin material, as seen in High Power Field. c. A free-floating rice body is composed of central pauci-cellular eosinophilic materials, surrounded by fibrin. d. High Power Field of the center of the rice body, without evidence of chondroid tissue or chondrocytes.



Post-operative follow-up at 24 months showed no recurrence of swelling. Patient was asymptomatic and shoulder function was normal with full power and motion range.

LITERATURE REVIEW PROCESS AND RESULTS

An English literature search was conducted in the PubMed database in the past 20 years, from 2001 to 2021. An initial search including “rice body” and “polymyositis” yielded no result. A second search was conducted using "rice body" AND ("bursa" OR "bursitis"). The abstracts were screened, and articles describing bursitis outside shoulder, infection, or lack of histological description, were excluded. Six case reports were found and listed in Table 11-6, including 6 females and 2 males. The majority of the case reports described multiple rice bodies of few millimeters in diameter, with histological findings of fibrin and inflammatory infiltrate, and were compatible with our findings. On the other hand, two case reports described the presence of osteochondral lesions, and two cases reported loose bodies of more than 1cm in diameter, which deviated from our current findings.

DISCUSSION

Multiple rice bodies are made of amorphous material, surrounded by collagen and fibrin. Although the exact etiology is unknown, the condition has been reported to be associated with tuberculosis and rheumatoid arthritis, and occasionally trauma and chronic bursitis[1]. The pathophysiology of rice body formation also remains unclear. Possible origins include micro-infarcted synovium leading to synovial shedding, and spontaneous formation in the synovial fluid with subsequent fibrin aggregation.

We reported the first case of spontaneous development of shoulder bursal rice bodies associated with polymyositis. Polymyositis is a rare disorder with unknown etiology, manifesting as symmetrical weakness of limb-girdle muscles, raised muscle enzymes and electromyographic evidence of myositis[7]. Non-musculoskeletal manifestations include dysphagia, arthritis, interstitial lung disease and association with malignancy. The association between polymyositis and rice bodies formation has never been reported. Polymyositis and rheumatoid arthritis have been regarded as distinct clinical syndromes[8], with little sharing of clinical features and serological markers. On the other hand, myositis has been known to have overlap syndromes with scleroderma, systemic lupus erythematosus, and mixed connective tissue disorder.

The diagnosis of rice bodies formation largely depends on radiological and histological findings. As non-calcified rice bodies will be invisible in X-ray, MRI is the imaging modality of choice. Typical findings include numerous filling defects inside fluid-filled bursa, being isointense or hypointense to skeletal muscles in both T1- and T2-weighted images[6]. For histological findings, rice bodies are composed of eosinophilic materials surrounded by fibrin and collagen. The cyst wall lining shows synovial hyperplasia with mild inflammatory infiltration.

Synovial chondromatosis is another main differential diagnosis for bursal inflammation with multiple loose bodies formation. While rice bodies are associated with inflammatory conditions, synovial chondromatosis is characterized by synovial metaplasia with chondrocytes formation. Consequently, cartilaginous masses can form in the subserous layer of the synovium, subsequently detaching from cyst lining to become loose bodies6. These pearly white loose bodies are usually larger and variable in size compared with rice bodies. Radiologically, these calcified bodies can be detected by plain radiographs, and will be evident as hyperintense signal in T2 MRI imaging. Histologically, loose bodies in synovial chondromatosis are

characterized by annular calcifications and irregularly distributed clusters of chrondrocytes within a hyaline matrix, forming cartilaginous or osteocartilaginous nodules. Table 2 summarizes the distinguishing features between rice bodies and synovial chondromatosis.

Both arthroscopic and open approaches have been advocated for treatment of bursal rice bodies[1-6]. One case of arthroscopic drainage developed recurrence, which was subsequently managed by aspiration and steroid injection. Whether the bursa needs to be partially debrided or completely resected remains inconclusive. Synovectomy, debridement or complete bursal excision procedures have been performed, with no recurrence up to 1 or 2 years. In the most recent literature, the author emphasized the prognostic importance to completely eradicating the "red" inflamed synovium using the Coblator device, which aids in ablating the inflamed tissue easily and completely[1].

CONCLUSION

Rice bodies formation in shoulder bursa has been rarely described in several case reports. The condition is found mostly associated with tuberculosis and rheumatoid arthritis. Our study is the first case report of rice body formation with polymyositis. MRI remains the imaging modality of choice. Histologically rice bodies are composed of eosinophilic cores surrounded by fibrin, and the absence of chondrocytes or osteochondral fragments distinguishes itself from synovial chondromatosis. Treatment includes arthroscopic synovial ablation or open bursectomy. Recurrence is unlikely after surgery.

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