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Case Report

Subscapularis pyomyositis presenting as shoulder stiffness mistaken as frozen shoulder in young female: a case report ☆,☆☆

*In Keun Park, MD, Suk-Hwan Jang, MD**

Department of Orthopedic Surgery, Inje University Seoul Paik Hospital, Mareunnae-ro 9, Jung-Gu, Seoul, South Korea

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ABSTRACT

Pyomyositis of the shoulder region in young healthy patients is rare but can lead to fatal overwhelming sepsis. Here we present the case of an otherwise healthy 32-year-old female patient with pain and stiffness in the right shoulder. Initial treatment with physical therapy and injection was ineffective. Magnetic resonance imaging of the right shoulder suggested subscapularis intramuscular sarcoma but excision of the muscle and biopsy revealed organized subscapularis pyomyositis. This case demonstrates the importance of investigating predisposing conditions in young patients with painful stiffness mimicking frozen shoulder that does not respond to nonoperative treatment.

Introduction

Adhesive capsulitis (frozen shoulder) is one of the most common causes of scapulohumeral pain and loss of motion. The condition develops in 2%-5% of the general population, but is especially prevalent in females aged 40-60 years [1,2]. Thus, in young female patients with painful loss of shoulder motion, the etiology may be difficult to determine, especially in the absence of a complete medical history [2].

While several conditions commonly lead to shoulder stiffness, pyomyositis is a rare cause. The estimated incidence of pyomyositis is 0.5 cases per 100,000 person-years, and only 8% of these patients present at the hospital with shoulder discomfort [3]. The diagnosis may also be missed in young patients without specific risk factors such as diabetes or an immunocompromised status [4]. However, failure to diagnose pyomyositis can lead to a catastrophic outcome, ranging from chronic osteomyelitis to death.

Herein, we present a rare case in which an otherwise healthy young female patient, presenting with pain and stiffness of the right shoulder and initially treated nonoperatively for primary frozen shoulder), was later shown to have pyomyositis of the rotator cuff.

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* Corresponding author.
E-mail address: orthopodjang@gmail.com (S.-H. Jang).
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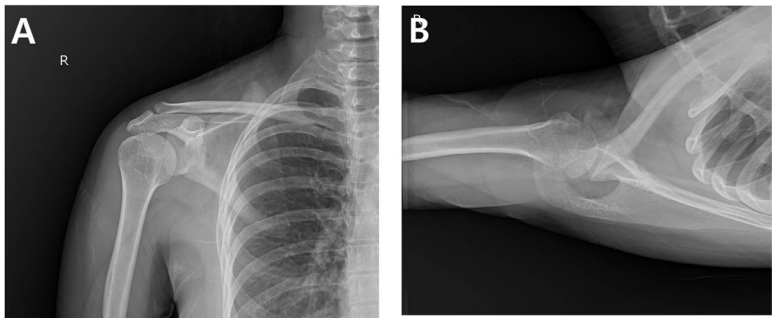

Fig. 1 – Initial plain radiographs with no abnormalities.

Case report

A 32-year-old female patient with pain and stiffness in the right shoulder presented to our hospital and was initially treated conservatively with medication, physical therapy, injections, and extracorporeal shockwave therapy for 6 months based on a diagnosis of frozen shoulder. She had no signs or symptoms of infection, and her laboratory test results were within the normal range (WBC, 9,740/mm^3; ESR 40 mm/h; CRP, 0.1 mg/dL). Consent for submission and publication of this case report including images and associated text has been obtained from the patient.

Plain radiographs showed no findings associated with abnormal pain or stiffness in a young patient (Fig. 1).

Physical examination revealed diffuse tenderness of the anterior shoulder. The range of motion was as follows: 100° of forward elevation, 10° of external rotation on 0° abduction, 30° of external rotation on 90° abduction, and internal rotation of the shoulder joint below the level of the buttocks. To alleviate the stiffness, steroid was injected into the glenohumeral joint via a posterior approach. Ultrasonography performed at that time revealed no evident pathology.

Despite intra-articular steroid injection, unlike in patients with common shoulder stiffness, there was no improvement in pain or stiffness at the 1-month follow up. Magnetic resonance imaging (MRI) was then performed to rule out mechanical obstruction of the joint that limited motion. The initial MRI findings of a well-defined multilobular mass with a definite septum suggested intramuscular sarcoma in the subscapularis (Fig. 2). Although intramuscular sarcoma in young female patients is very rare, it seemed more likely than pyomyositis because the patient had no symptoms related to infection, only mechanical discomfort.

According to surgical guidelines for patients with suspected sarcoma, radical excision of the subscapularis muscle was performed and the tissue was sent for pathological evaluation. During surgery on the mass, diffuse fibrotic tissues covering the rotator interval were noted and may have been the cause of the restricted shoulder motion. However, the histopathologic report concluded that the subscapularis intramuscular mass lesion was mainly composed of atrophic skeletal muscle and fibrotic tissues with abscess, which are characteristic features of pyomyositis.(Fig. 3). The abscess measured 4.0 × 4.0 × 2.5 cm. A culture taken from the lesion was negative.

At the 1-year follow up, the patient was in good condition without any symptoms. Physical and radiological examinations revealed no evidence of recurrence.

Discussion

Adhesive capsulitis of the shoulder, described by Codman in 1934 as “frozen shoulder” to emphasize the debilitating loss of motion, is one of the most common conditions encountered by orthopedic surgeons. The pathological process involves the formation of excessive scar tissue or adhesions around the glenohumeral joint, resulting in stiffness and pain in the shoulder region [5]. Despite the high prevalence of frozen shoulder and the many attempts to determine its natural history, it remains poorly understood. Hence, diagnosis may be difficult, especially in young in patients who present to the hospital with unexplained stiffness of the shoulder.

Adhesive capsulitis is usually classified as primary or secondary. Primary (or idiopathic) adhesive capsulitis can occur spontaneously without any specific catalyzing event, whereas secondary adhesive capsulitis often follows severe trauma around the shoulder joint, such as fracture dislocation of the glenohumeral joint [6].

The risk factors for frozen shoulder include female sex, previous history of trauma, HLA-B27 positivity, age >40 years, history of medical disease and prolonged immobilization of the glenohumeral joint. The incidence of adhesive capsulitis in the general population is 3%-5%, but is as high as 20% in patients with diabetes. Among patients with adhesive shoulder capsulitis, 70% are women [7,8]. Demographic studies have shown that most patients with adhesive capsulitis (84.4%) are aged between 40 and 59 years [9].

Based on this information, adhesive shoulder capsulitis would have been unusual in our patient, who is a healthy young female with an unremarkable medical history. Her case shows that in patients with unexplained stiffness and pain, infection should be considered.

Primary pyomyositis is a rare, subacute, primary, and deep muscle infection thought to result from transient bacteremia, rather than a contagious skin, bone or soft-tissue infection.

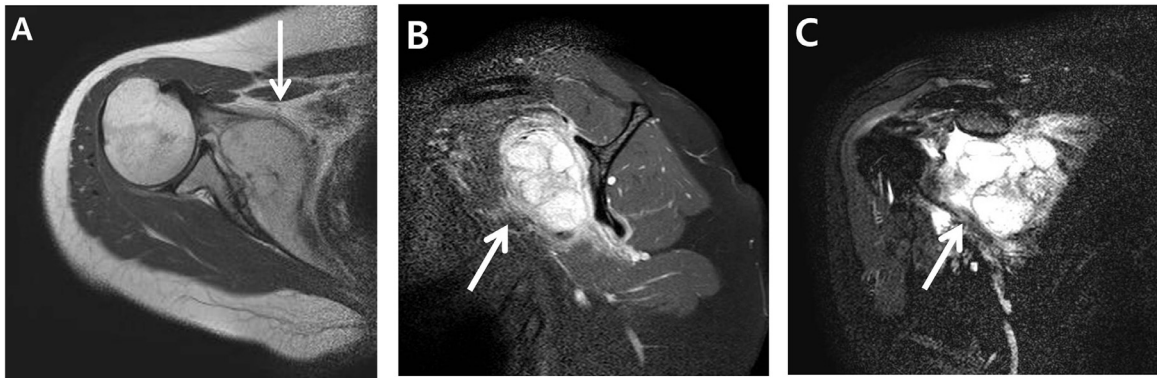

Fig. 2 – T1 Axial (A), T2 coronal (B), and T1 gadolinium enhanced fat-suppressed sequence sagittal, (C) 1.5T MRI images of right shoulder show 4.5 x 4.4 x 2.9cm sized well-defined lobulated mass in subscapularis muscle, and multiple septa-like structure in it (T1WI: low SI, T2WI: heterogeneous high SI).

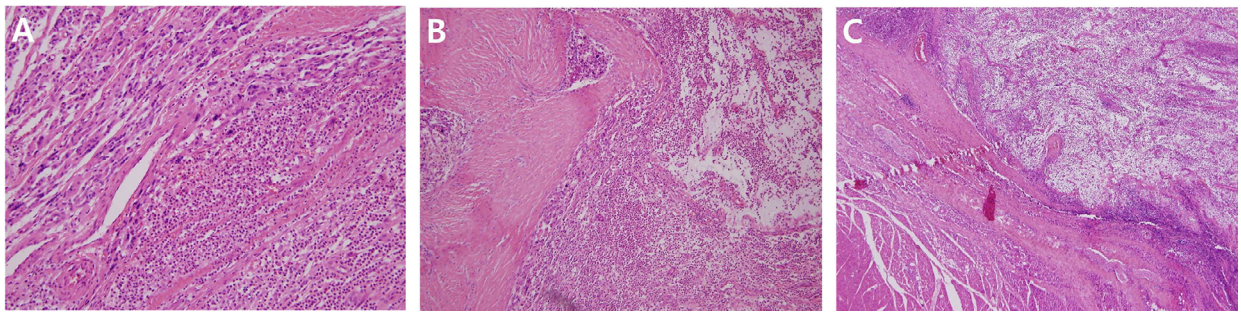

Fig. 3 – (A) Histopathology shows the atrophic skeletal muscle with abscess (H&E x 100), (B) Fibrosis with abscess (H&E x 100), (C) inflammatory infiltrates containing many granulated cells (H&E x 40).

The quadriceps, gluteal, and iliopsoas muscles are the most commonly affected areas, whereas involvement of the muscles of the shoulder or upper arm region is seen in <10% of pyomyositis cases [3]. In many patients, pyomyositis manifests as a localized abscess or diffuse inflammation around the lesion, which may lead to mechanical irritation. *Staphylococcus aureus* and *Streptococcus pyogenes* are the most common causative organisms, with positive culture rates from blood samples and needle aspirations of 31% and 70%-90%, respectively [10]

Although immunocompromised patients in tropical climates are more susceptible to infection by organisms causing pyomyositis, in immunocompetent patients in temperate climates 75% of infections are caused by *S. aureus*. In those cases, the patients are often younger and have a history of either trauma or injury during active exercise, or an underlying dermatologic disorder [11]. The pathophysiology of pyomyositis may thus involve an undiagnosed injury to the surrounding muscle tissue, which increased the blood supply to the damaged tissue and thus altered the normally biologically resistant environment, thereby making the patient vulnerable to infectious agents.

Three stages of pyomyositis are described in the literature: invasion, suppuration, and sepsis [12]. During the first stage, patients usually complain of dull, cramping pain in the affected region and mild fever. If the diagnosis is made promptly at this stage, antibiotic treatment based on culture is usually effective. Unfortunately, as in this case, in 90% of patients the diagnosis is made during the suppurative or purulent stage (1-3 weeks after the onset), by which time an abscess has already formed in the muscle. Treatment at these stages consists of completely excising the abscess and aggressive antibiotic therapy.

As this case demonstrates, in young patients with painful stiffness mimicking frozen shoulder that does not respond to nonoperative treatment, predisposing conditions should be carefully investigated. In our patient, removal of the subscapularis muscle proved unnecessarily radical. However, because a tumor was initially suspected, this was considered the appropriate therapeutic strategy. The exact pathology of the pyomyositis in our patient was unclear and both its clinical manifestation and MRI findings hindered correct diagnosis during the early stage. However, an inflammatory reaction caused by a subscapularis lesion might have affected the whole shoulder joint capsule, resulting in abnormal stiffness.

In conclusion, unusual stiffness and pain in the shoulder of a young female patients suggests a wide range of disease entities, from simple frozen shoulder to (albeit rarely) infection. Thorough early assessment, including laboratory tests

and specific imaging, may prevent inappropriate and delayed treatment.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2020.08.006.

REFERENCES

[1] Kelley MJ, Shaffer MA, Kuhn JE, Michener LA, Seitz AL, Uhl TL, et al. Shoulder pain and mobility deficits: adhesive capsulitis. J Orthop Sports Phys Ther 2013;43(5):A1–31.

[2] Ryan V, Brown H, Minns Lowe CJ, Lewis JS. The pathophysiology associated with primary (idiopathic) frozen shoulder: a systematic review. BMC Musculoskelet Disord 2016;17(1):340.

[3] Corey SA, Agger WA, Saterbak AT. Acromioclavicular septic arthritis and sternoclavicular septic arthritis with contiguous pyomyositis. Clin Orthop Surg 2015;7(1):131–4.

[4] Burdette SD, Watkins RR, Wong KK, Mathew SD, Martin DJ, Markert RJ. Staphylococcus aureus pyomyositis compared with non-Staphylococcus aureus pyomyositis. J Infect 2012;64(5):507–12.

[5] Neviaser AS, Neviaser RJ. Adhesive capsulitis of the shoulder. J Am Acad Orthop Surg 2011;19(9):536–42.

[6] McAlister I, Sems SA. Arthrofibrosis after periarticular fracture fixation. Orthop Clin North Am 2016;47(2):345–55.

[7] Sheridan MA, Hannafin JA. Upper extremity: emphasis on frozen shoulder. Orthop Clin North Am 2006;37(4):531–9.

[8] Manske RC, Prohaska D. Diagnosis and management of adhesive capsulitis. Curr Rev Musculoskelet Med 2008;1(3-4):180–9.

[9] Boyle-Walker KL, Gabard DL, Bietsch E, Masek-VanArsdale DM, Robinson BL. A profile of patients with adhesive capsulitis. J Hand Ther 1997;10(3):222–8.

[10] Christin L, Sarosi GA. Pyomyositis in North America: case reports and review. Clin Infect Dis 1992;15(4):668–677.

[11] Block AA, Marshall C, Ratcliffe A, Athan E. Staphylococcal pyomyositis in a temperate region: epidemiology and modern management. Med J Aust 2008;189(6):323–5.

[12] Bickels J, Ben-Sira L, Kessler A, Wientroub S. Primary pyomyositis. J Bone Joint Surg. 2002;84(12):2277–86.