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Case Report

Subscapular bursitis as a rare manifestation of dermatomyositis: a case report

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Abstract

Dermatomyositis (DM) is characterized by proximal muscle weakness and characteristic skin rash. Pain is a less common feature and usually indicates inflammation of extramuscular structures such as fascia. Here we report a rare case of subscapular bursitis in a 48-year-old woman with DM. She initially presented with severe, sharp, stabbing pain in her right shoulder that worsened with shoulder movement. Magnetic resonance imaging (MRI) revealed inflammation in the right subscapular bursae. A few months later, the patient developed periungual erythema, Gottron's papules, and shawl sign with muscle pain in her thighs. DM was diagnosed based on the presence of interface dermatitis on skin biopsy and diffuse muscle inflammation on MRI. Bursitis and myalgia responded incompletely to nonsteroidal anti-inflammatory drugs but promptly to corticosteroids. Here we report a case of subscapular bursitis as a rare manifestation of DM. Pain in patients with DM may warrant physicians to evaluate for the presence of additional inflammatory processes in the perimuscular structures.

Keywords: Dermatomyositis, bursitis, pain

Introduction

Dermatomyositis (DM) is a chronic autoimmune disease involving muscles and skin as the main target of inflammation (1). While proximal muscle weakness is the key musculoskeletal feature, pain is not common in idiopathic inflammatory myopathy. Therefore, pain often indicates involvement of extramuscular structures. Bursitis, inflammation of the bursae, is painful and extremely rare in DM (2, 3). Here we report a unique case of subscapular bursitis as the initial presentation of DM.

Case presentation

A 48-year-old woman presented with a 5-day history of stabbing right upper back pain. Her pain was precipitated by active shoulder movement and improved with rest. The patient denied any preceding trauma or unusual exercise. On examination, the area delimited by her right scapula was tender to pressure. Range of shoulder motion was not limited in any directions. Proximal and distal muscle strength was fully preserved. Facial erythema, involving the bridge of her nose and nasolabial folds, was noted (Figure 1a). The patient's upper arms revealed an ulcerating maculopapular rash on erythematous background with prominent scar formation (Figure 1b). C-reactive protein levels were elevated to 11.17 mg/dL (normal <0.05 mg/dL), and the erythrocyte sedimentation rate increased to 64 mm/h (normal <20 mm/h). Anti-nuclear antibodies were detectable at 1:40. Antibodies to extractable nuclear antigens (ENA) were not identified. Rheumatoid factor was positive at 185 IU/mL (normal <14 IU/mL), anti-cyclic citrullinated peptide (CCP) antibodies were undetectable. Magnetic resonance imaging (MRI) of the right shoulder showed scapulothoracic bursitis (Figure 2a). Skin biopsies obtained from face and left arm revealed perivascular, perifollicular lymphocytic infiltrates in the superficial dermis, suggestive of interface dermatitis. Treatment for subscapular bursitis with nonsteroidal anti-inflammatory drugs (NSAID) was initiated. Minocycline and corticosteroids were used topically for the skin rash. While the facial rash improved with therapy, the patient's subscapular bursitis worsened. The ulcerating rash on her arms persisted. On follow-up, ultrasonography confirmed a hypoechoic lesion measuring 2.3 cm in diameter adjacent to the serratus anterior muscle. Attempted aspiration of the bursae failed. Dramatic improvement of subscalpular pain was finally achieved with prednisolone at 20 mg/day for 1 week.

Over the following 3 months, the patient developed periungual erythema, Gottron's papules, and shawl sign (Figure 1c) and started to complain of muscle discomfort in both legs. Proximal muscle strength in her right thigh was decreased (Grade IV-/V). Other proximal and distal muscle groups were intact. Creatine phosphoki-

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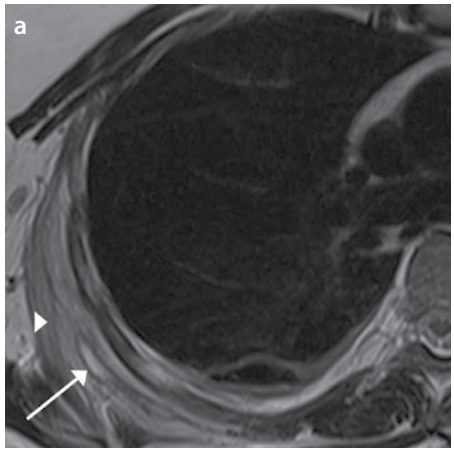
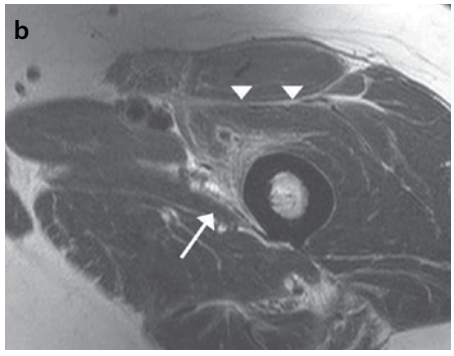
Figure 1. a-c. Erythematous facial rash that did not spare the nasolabial folds (a), maculopapular dermal lesions with shallow ulcers on the patient's upper arms (b), and erythematous rash on the upper back (shawl sign) (c)





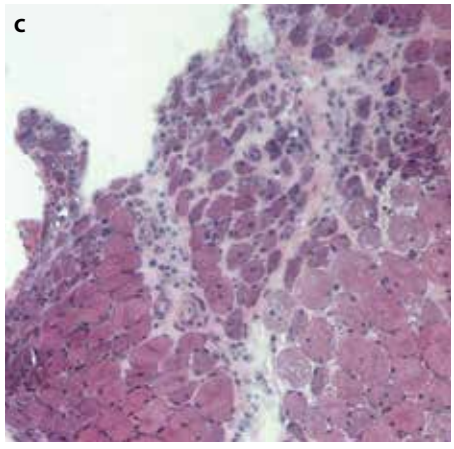


Figure 2. a-c. T2-weighted MRI image (a) of the patient's right shoulder and the posterior chest wall demonstrate a hyperintense, fluid-filled structure (arrow) and edema in the adjacent serratus and latissimus muscle (arrowhead), compatible with the clinical diagnosis of subscapular bursitis. Axial T2-weighted MR image of the left thigh muscle (b) showed diffuse T2-hyperintensities of the deep fascial planes (arrowheads) and adjacent muscle edema (arrow), were observed, indicative of fasciitis and myositis in DM. Excisional biopsy of the left quadriceps femoris muscle (c) revealed perifascicular atrophic myofibers with few inflammatory cells. Original magnification ×10

nase levels were normal at 59 IU/L. Aldolase was elevated at 9.0 U/L (normal <7.6 U/L). MRI of the thighs revealed diffuse inflammation along musculofascial planes with muscle edema bilaterally (Figure 2b). Muscle biopsy showed evidence of degenerating and regenerating myofibers, and perifascicular atrophy with few inflammatory cells (Figure 2c). No malignancy was detected. A diagnosis of DM was made and therapy with high-dose corticosteroids initiated. This was followed by prompt improvement of the patient's her myalgia. The ulcerating skin rash improved slowly with therapy.

Discussion

While muscle weakness is the key feature of the idiopathic inflammatory myopathies (IIM), myalgia is less common in patients with myositis (1). The presence of profound myalgia may indicate additional inflammatory processes involving fasciae and bursae (4, 5). To the best of our knowledge, we report the third case of DM-associated bursitis, which in this case preceded the development of muscle weakness by months and responded well to corticosteroid therapy (2, 3).

Bursae are fluid-filled sacs underlying tendons or muscles at bony prominences to reduce friction. Bursitis often results from overuse injury with repetitive motion, leading to microtears and subsequent inflammation. Bursitis can occur alone or in association with other diseases including infection, trauma, rheumatoid arthritis, and systemic sclerosis (6). The prevalence of bursitis associated with DM is unknown. Bursitis in DM may be underappreciated as associated pain is more often attributed to myositis and fasciitis. Similar to polymyalgia rheumatica, which causes subjective myalgia due to bursitis in the absence of myositis, pain associated with myositis may be independent of muscular inflammation (4). Indeed, isolated fasciitis is often associated with muscle pain and weakness (5). A recent report by Yoshida et al showed a high prevalence of fasciitis in DM, initially believed to be rare in IIM (7). Fasciitis may in fact represent an early lesion in the evolution of DM. In line with this, the patient described here developed bursitis prior to the onset of muscle symptoms.

The rarity of bursitis in DM may suggest that a presentation with both conditions is coincidental. However, DM may increase the development of bursitis by several mechanisms. Soft tissue (micro) calcification might cause mechanical trauma and microtears (8). Moreover, systemic vasculopathy with endothelial dysfunction observed in DM may lead to hypoxic damage of the thin bursae, which has limited vascular supply. Immune complex deposition in the synovial layer of bursae may fix complement and precipitate bursitis (9). The persistence and poor response of bursitis to NSAIDs in this patient suggests that mechanisms underlying the development of bursitis may indeed be distinct from common overuse injury, which typically responds well to NSAIDs (10). Rather, bursae inflammation in this patient may be mediated by autoimmune processes, similar to the synovitis observed in

rheumatoid arthritis, which responds poorly to NSAID therapy. Severe pain in patients with IIM may indicate additional inflammatory processes unrelated to muscle inflammation including bursitis or fasciitis.

To summarize, we report a rare case of subscapular bursitis as the initial presentation of DM. Further study is necessary to establish the incidence of bursitis in DM, especially in IIM patients presenting with musculoskeletal pain.

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