Multiple Oleomas Presenting as Dermatomyositis

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The injection of silicone or oil-based substances into muscles for cosmetic reasons can result in masses called oleomas. We present the case of a 38 year old man who presented with muscle pain and swelling associated with a high C-reactive protein (CRP). This was initially thought to be due to an idiopathic inflammatory muscle disease. However, detailed questioning and subsequent biopsy revealed other possibilities.

PATIENT DESCRIPTION

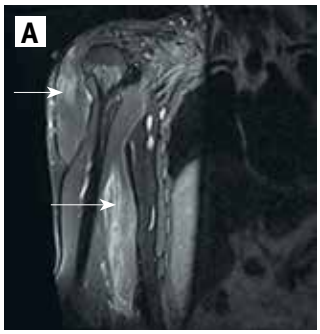
A 38 year old man originating from Qatar presented to our unit with a history of dermatomyositis that had been resistant to treatment despite the use of prednisolone and azathioprine. Two years earlier he experienced symmetric muscle pain and swelling in the upper limbs. There was also a history of some skin peeling around the dorsal surface of the hands and as a result he had undergone a number of investigations. These had shown an acute-phase response and a mild normochromic normocytic anemia (hemoglobin 12 g/dl), but creatinine and liver function tests were normal. In particular, creatinine phosphokinase (CPK) was also normal. In addition, rheumatoid factor (RF) and antinuclear antibodies (ANA) were negative; blood cultures, urine culture and chest X-ray were normal. A magnetic resonance imaging scan (MRI) with contrast of both upper extremities showed evidence of active myositis especially in the deltoid, biceps and triceps muscles. A subsequent biopsy taken from an area of deep tenderness and swelling in the region of the left deltoid muscle was described as showing minimal non-specific inflammation; Congo red staining to seek amyloid was negative.

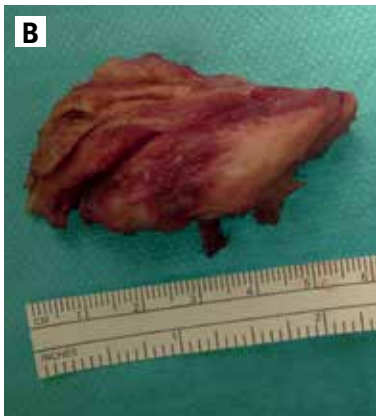
Based on the patients' history and presentation and these investigations, a diagnosis of dermatomyositis (DM) was made. Treatment was started with prednisolone and azathioprine (AZA). The dose of prednisolone decreased gradually from 60 to 40 mg per day and AZA increased gradually from 50 to 200 mg per day (approximately 2.5 mg/kg/day). Despite this there was no apparent improvement after 5 months and the patient stopped treatment with no significant deterioration.

At his latest presentation in our facility the patient complained mainly of symmetric pain and swelling in a number of areas including the triceps, biceps and deltoid muscle groups. On examination, these areas appeared tender and swollen with a palpable increase in temperature. The involved sites appeared as discrete hard swellings but the associated inflammatory tissue did not appear to extend through all the muscles involved. Muscle strength was normal in all muscle groups. Otherwise, the patient appeared well and further examination for features of myositis and connective tissue disease was unremarkable. In addition, from the history and examination there was no suspicion of underlying malignancy.

Further investigations showed hemoglobin 147 g/L (130–17 g/L), white blood cells (WBC) 9.2 x10^9/L (4.0–10.0), slightly raised eosinophils 0.6 x10^9/L (0.02–0.5), high erythrocyte sedimentation rate (ESR) 56 mm/hour, raised CRP 129 mg/L, and normal CPK 89 IU/L (26–140). Renal function, thyroid function and glucose were all normal. Serum angiotensin-converting enzyme (ACE) was slightly raised at 66 U/L (8–52), as was gammaglobulin, 41 g/L (20–40). Autoantibodies including ANA, extractable nuclear antigens (ENA) and RF were all negative, anti-double stranded DNA antibodies were 7 IU/ml (< 50), IgG anticardiolipin antibodies 4.6 U/ml (0–17) and IgM anticardiolipin antibodies 1.5 U/ml (0–15), but lupus anticoagulant screening was positive. The complement factor-3 (C3) and complement factor 4 (C4) were both raised: 2.36 g/L (0.9–1.8) and 0.59 g/L (0.1–0.4) respectively. A QuantiFERON test for tuberculosis (TB) was negative. Urine analysis was normal and urine culture showed no growth. A chest X-ray was normal. MRI of the musculature of the upper body revealed evidence of inflammatory changes in discrete patches within biceps, triceps and deltoid bilaterally with hyperintense signal on T1-weighted images and heterogeneous intensity on T2 [Figure 1A].

A plastic surgeon performed a deep biopsy of a lesion within the right triceps muscle [Figure 1B], which on histological examination showed areas of homogenous material consistent with a foreign body, such as an oil, surrounded by a granulomatous reaction [Figure 1C]. These findings led to further direct questioning of the patient who reported having received several injections of an oily substance into his muscles for cosmetic reasons in an attempt to enhance their definition. He did not experience any problems for about 4 years after receiving the last injection. However, pain and swelling had developed and continued for 2 years prior to review in our unit. The pain and swelling had become more acute in the past 3 months.





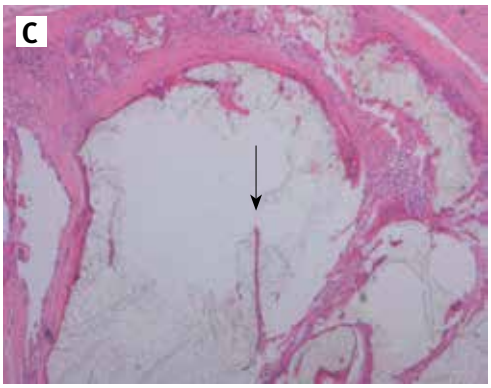


Figure 1. [A] MRI T2-weighted image of upper extremity showing symmetric hyper-intense STIR signal suggestive of inflammation within the deltoid, biceps and triceps muscles

[B] Muscle specimen from excision biopsy of the right triceps muscle

[C] Histopathology of muscle specimen (hematoxylin and eosin stain) with a foreign body-type reaction consistent with a granulomatous reaction to lipid

COMMENT

The injection of silicone or oil-based substances including paraffin, sesame oil, walnut oil and purified long-and medium-chain emulsions has been described a number of times although most reports appear in the non-English language literature [1]. Masses associated with this practice are commonly called oleomas, paraffinomas or lipogranulomas [2]. The most common presentation is painful muscle fibrosis. In the long term, side effects may include disfiguring scarring, loss of function, and deformity. Also reported are the side effects associated with such injections, such as infections, ulcers, pruritus, oil embolism, myocardial infarction, pulmonary and neurological complications, with other rarely described complications including severe hypercalcemia and nephrocalcinosis [3]. Rhabdomyolysis has been documented following injection of anabolic androgenic steroids in bodybuilders [4]. There are very few reports on how to treat these lesions, although the use of a number of medications including antibiotics have been described (long-term oral tetracycline, corticosteroids, colchicine, compression bandaging, surgical drainage and plastic surgery) [5].

For our patient we had a number of concerns. These included his ongoing pain, deformity and reduced muscle function. We also considered the possibility that with a persistently raised CRP of more than 100 mg/L secondary amyloidosis might develop. With this in mind the following treatment regimen was begun. We surgically removed as much inflamed and foreign tissue as possible while preserving muscle function. Six areas were targeted in a staged procedure to remove inflammatory masses from the right and left deltoid, biceps and triceps muscles. The rationale behind this was to attempt to "debulk" as much foreign material as possible with the hope that it would reduce the inflammatory response to the foreign material. The followed protocol was then started: prednisolone 7 mg daily, colchicine 500 µg twice a day, methotrexate 10 mg weekly (to be titrated up), and folic acid 5 mg weekly. We also planned an amyloid (SAP) scan to look for amyloid deposits.

This case highlights the dangers of unregulated cosmetic procedures and the powerful effect of long-term exposure to a foreign antigen. In addition, it illustrates the possibility that foreign body injection into muscle groups to enhance definition may lead to the mistaken diagnosis of inflammatory myositis. The key to the diagnosis was of course the history of muscle injection, but it is perhaps understandable that the patient was reluctant to divulge this information. The patient's reaction to the injected substance was also interesting. When we reviewed the situation there was an intense foreign body response with an elevated CRP but normal CPK. The serum ACE was also raised, presumably due to the granulomas evident on the biopsy. However, the patient only developed the reaction 4 years after the injections, suggesting a loss of tolerance. We questioned him about other factors that may have led to a loss of tolerance, such as immunization, infection or other surgical procedures, but there were none. We hope that debulking the foreign bodies and modest immunosuppression will preclude further pain, muscle damage and the negative consequences of a long-term uncontrolled inflammatory response.

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