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Iatrogenic flexor tendon rupture caused by misdiagnosis of sarcoidosis related flexor tendon contracture as tenosynovitis: A case report

Misdiagnosis of sarcoid related finger contracture

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Abstract

BACKGROUND

Sarcoidosis is a multi-system disease characterized by granuloma formation in various organs. Sarcoidosis related flexor tendon contracture is uncommon in clinical settings. This contracture is similar to that of stenosing tenosynovitis, potentially leading to misdiagnosis and mistreatment.

CASE SUMMARY

We describe a rare experience of the treatment of a patient with finger flexion contracture of her right ring and middle fingers who had been misdiagnosed as tenosynovitis, and received acupotomy release of the A1 pulley of her middle finger that resulted in iatrogenic rupture of both superfical and profundus flexors (FDS & FDP). Radiological presentation then showed multiple sarcoid involvement in pulmonary locations and the ipsilateral forearm as well. A diagnosis of sarcoidosis was made given the presence of noncaseating granuloma with tubercles consisting of Langhans giant cells with infiltration of lymphocytes in biopsy. The patient suffered another following spontaneous rupture of the repaired tendon of the middle finger. Satisfactory results were achieved at a follow-up of 10 mo after reoperation.

CONCLUSION

Sarcoidosis with the involvement of musculoskeletal system leading to finger contracture is rare, thus great caution should be paid when dealing with such patients to avoid wrong treatment.

Key Words: Sarcoidosis; Finger flexor contracture; Iatrogenic injury; Misdiagnosis; Granulomatous myopathy; Case report

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Core Tip: Contracture of fingers due to tenosynovitis is frequently encountered in clinical settings. However, misdiagnosis could ensue when some rare causes such as sarcoidosis are neglected. We report a case where such a misdiagnosis occurred and led to iatrogenic injury. The mainstream treatment method for contractures related to sarcoidosis is surgical excision of muscular lesions with varied outcomes in several case reports. In our case, considering the contracture lesion at flexor digitorum profundus and the initial iatrogenic tendon rupture, cross lengthening of flexor digitorum superficialis and profundus was attempted to restore flexion, and achieved satisfactory results.

INTRODUCTION

Granulomatous involvement of skeletal muscle (also known as granulomatous myopathy) is common in patients with sarcoidosis and most are asymptomatic[1]. These patients suffering finger flexion contracture are extremely rare. To our best knowledge, only several cases of such contractures have been reported in English literature[2–8].

Contracture of finger flexors is frequently seen in tenosynovitis (also known as trigger finger) in clinical settings.[9] Trigger finger is caused by inflammation and constriction of the retinacular sheath through which the flexor tendons run as they pass from the palm of the hand into the finger, leading to pain and movement restriction. The management of tenosynovitis has been well established, ranging from conservative to surgical treatment.[9–11] Since finger contracture of other causes such as sarcoidosis is rare, misdiagnosis of tenosynovitis could happen, leading to empirical treatment with poor outcomes or severe complications.

In this case report, we present a patient with finger contractures was misdiagnosed as having trigger finger, and received acupotomy release[12,13] (similar to a percutaneous release), resulting in iatrogenic rupture of both superfical and profundus flexors of her right middle finger. In addition, a systematic literature review regarding finger contractures caused by sarcoidosis was also carried out.

CASE PRESENTATION

Chief complaints

A 44-year-old healthy woman presented to our hand surgery department with spontaneous flexor tendon rupture of her right middle finger and contracture of her right ring finger.

History of present illness

She had been diagnosed as tenosynovitis of the right ring finger and middle finger in a local hospital, and received acupotomy release of her middle finger 2 wk prior to admission. Five days after the release, a sudden complete loss of active flexion of her right middle finger was noticed during routine domestic activity. Two months after operation, rerupture of the repaired tendon of the middle finger was encountered when the patient was pulling on her pants.

History of past illness

The patient denied any past illnesses.

Personal and family history

The patient denied any personal or family history of related diseases.

Physical examination

Loss of active flexion of the right middle finger and contracture of the right ring finger was discovered. There was no swelling, tenderness or numbness at the digits. No other positive findings were revealed during systematic physical examination. During operation, entire lacerations of both the flexor digitorum superficialis (FDS) and flexor digitorum profundus (FDP) of the right middle finger were identified at Zone II around the MP joint (Figure 1). Significantly, high level of tension and a loss of elasticity were observed in the proximal end of FDP, while the proximal FDS seemed to be normal in tension and elasticity.

Laboratory examinations

Initial laboratory studies revealed high alkaline phosphatase (116U/L, 35~100), serum creatinine, creatine phosphokinase (CPK) and calcium levels were normal. No further positive findings were noted in laboratory examinations.

Histopathology and HE staining revealed granulomatous involvement in the FDP muscles and tendon, showing a typical sarcoid with tubercles that consists of Langhans' giant cells and lymphocytes (Figure 3 a, b).

Imaging examinations

High-resolution computed tomographic section showed widespread nodules in both lungs (Figure 4).

Magnetic resonance imaging (MRI) revealed in the right forearm muscles signs of a central dark star surrounded by a peripheral high signal area on axial images, and a

three-stripe pattern on coronal and sagittal images, typical of sarcoid myopathy (Figure 5).[14–16]

FINAL DIAGNOSIS

The patient was diagnosed with sarcoidosis related flexor tendon contracture and iatrogenic flexor tendon rupture.

TREATMENT

In the first surgical exploration, the distal FDP of the middle finger was anastomosed with the proximal end of FDS to restore its flexion function. Surprisingly, the ring finger remained in contracture after complete open release of the A1 pulley, indicating the contracture was not due to the tenosynovitis. No further intervention was performed after discussion with the patient intraoperatively. Routine postoperative management was carried out including splinting for 4 wk and corresponding rehabilitation.

Unfortunately, 2 mo after operation, re-rupture of the repaired tendon was encountered when the patient was pulling on her pants (Figure 2 a). Reoperation was carried out on her right middle and ring finger. Anastomoses of proximal FDS to the distal FDP using tendon grafting was performed for the middle finger, and direct repair was performed for the ring finger (Figure 2 b).

OUTCOME AND FOLLOW-UP

The patient partially regained motion of her middle finger, while contracture of her right ring finger remained after the first surgery attempt. After reoperation, she could use the hand without discomfort, despite a persistent mild stiffness at a follow up of 10 mo (Figure 2 c, d).

DISCUSSION

In our case, surgery was performed as the patient was presented to us with an iatrogenic rupture. Sarcoidosis was not considered until the second surgery attempt,

and was proven after MRI and chest CT imaging. After rerupture, a comprehensive medical history taking revealed an additional complaint of mild discomfort with tenderness in physical examination around the forearm flexor muscles, indicating involvement of muscular lesion. Furthermore, tenosynovitis should have been ruled out since the patient denied ever suffering pain or triggering around the metacarpophalangeal (MCP) joint, or had a history of aggravation after housework.

Despite the fact that contractures due to sarcoidosis is extremely rare, neglect of these distinct symptoms, and the absence of a systematic examination resulted in the misdiagnosis and the ensuing mistreatment.

The diagnosis of sarcoidosis is not well established but is recommended according to three major aspects: a compatible clinical and/or radiological presentation, the histological evidence of non-necrotizing granulomatous inflammation in one or more tissues and the exclusion of alternative causes of granulomatous disease.[17]

Chest radiograph has been the cornerstone of diagnosis of sarcoidosis since 1961, when Scadding proposed a standardized staging system.[18] Currently, computed tomography (CT) represents the reference standard for the assessment of both pulmonary findings and mediastinal lymph nodes.[19] In our case, MRI and biopsy results revealed lesions at forearm muscles, while a clear diagnosis of sarcoidosis was only made after CT imaging.

The treatment of sarcoidosis related contracture has been presented in several case reports with varied results.[2–7,20,21](Table 1)

Acute sarcoid myopathy improves under systemic treatment in most patients, while patients with chronic sarcoid myopathy commonly suffer severe disability such as flexor contractures and seldom recover after corticosteroid treatment, immunosuppressive or anti-tumor necrosis factor (TNF)-α.[22,23] In such chronic cases with finger contractures, surgical intervention has been attempted.[2,4,6,7] In 1996, Kazuo Ikeda *et al* held that contracture due to sarcoidosis could be successfully treated with surgical excision of lesion parts[4]. But in 2009, he restated that even with complete excision of the granulomatous lesion, a new lesion may appear in a previously healthy

area, so a radical cure by operation alone is difficult.[6] Conservative treatment has also been reported. Walter MC *et al* reported a case where the patient's contractures had improved after 9 mo's treatment of low-dose thalidomide.[21] And Bowers reported another patient aged 14 who improved under occupational therapy.[5] Notably, while in previous cases biopsy tests were focused on forearm muscles, our biopsy results revealed lesion at the tendon as well.

Coincidentally, Motomiya *et al*[7] also reported a patient with sarcoidosis related flexor contracture was initially misdiagnosed as trigger finger and received open surgery. Luckily, the tendon was fully exposed during operation and the misdiagnosis of tenosynovitis was then realized. In our case, the patient was misdiagnosed as tenosynovitis and was treated with acupotomy (a kind of blind release) at the first visit in a local hospital. The operation was not able to give further information about the contracture, inevitably resulting in iatrogenic flexor rupture when pursuing complete finger flexion release. Therefore, treatment of tenosynovitis using acupotomy should be of great caution in preoperative diagnosis.

In our case, due to the initial iatrogenic flexor tendon rupture, tendon grafting was performed to repair the FDP tendon, connecting the proximal end of FDS to the distal FDP. Notably, open surgery at the digit revealed normal tension at the FDS, and the FDS muscle was later proven to be free of lesion by biopsy results, thus this surgery was able to partially restore flexion to the affected fingers. In conclusion, this method was adapted due to the initial attempt to repair the tendon rupture, and achieved satisfying results to alleviate the contractures at a follow-up of 10 mo.

CONCLUSION

Sarcoid involvement of musculoskeletal systems causing finger contractures is rare, which could lead to misdiagnosis if clinicians are not aware. Our case shows that finger contracture release with connecting the normal proximal end of FDS to the distal FDP may be an option for such cases, though the management of sarcoidosis related flexor contracture is still controversial with varied outcomes based on literature review.

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