

Sarcoid tenosynovitis, rare presentation of a common disease. Case report and literature review

Zeid Al-Ani^{1*}, Teik Chooi Oh², Elizabeth Macphie³, Michael J Woodruff⁴

1. Northwest Radiology Training Scheme, Royal Preston Hospital, Preston, UK

2. Radiology Department, Royal Preston Hospital, Preston, UK

3. Rheumatology Department, Minerva Health Centre, Lancashire Care NHS Foundation Trust, Preston, UK

4. Trauma and Orthopaedics Department, Royal Preston Hospital, Preston, UK

* **Correspondence:** Zeid Al-Ani, Northwest Radiology Training Scheme, Royal Preston Hospital, Preston, UK
(✉ Zeid82@yahoo.com)

Radiology Case. 2015 Aug; 9(8):16-23 :: DOI: 10.3941/jrcr.v9i8.2311

ABSTRACT

Sarcoidosis is an idiopathic inflammatory disorder characterized by the presence of non-caseating tissue granulomas most commonly affecting lungs, lymph nodes and skin. Sarcoid skeletal involvement is relatively uncommon and in particular tenosynovitis. We describe an unusual case of sarcoidosis presenting with granulomatous tenosynovitis as the only manifestation of the disease, illustrating the radiological findings on different modalities followed by a review of the literature. Radiologists and clinicians should be aware of tenosynovitis as a manifestation of sarcoidosis as early and therefore appropriate treatment significantly alters patient's outcome and prognosis.

CASE REPORT

CASE REPORT

A 42 year old male was referred to the rheumatologist after developing a painless lump affecting the pulp of his right middle finger. He had been troubled for some years with swelling affecting his left index and middle fingers. His past medical history is only significant for hypercholesterolemia for which he is on Simvastatin 40 mg once daily. He had no other symptoms of note, specifically no respiratory symptoms and no history of any skin rashes or psoriasis. There was no family history of inflammatory arthritis or psoriasis and no recent travel history of note.

Examination showed fusiform, non-tender swellings affecting the left index and middle fingers with reduction in flexion at proximal interphalangeal (PIP) and distal interphalangeal (DIP) joints. He also had non-tender swelling of the pulp of his right middle finger. Examination of the remaining joints, chest, abdomen, cardiovascular system and skin did not show significant abnormalities. Routine blood

tests including inflammatory markers, full blood count, bone profile, renal and liver function tests were all normal. Angiotensin Converting Enzyme level was elevated at 92 microgram/L (normal 15-55).

Imaging findings

Bilateral hands radiographs showed fusiform soft tissue swelling around the left index and middle fingers phalanges and right middle finger distal phalanx but no bone involvement or cortical destruction (Figure 1).

Ultrasound examination of the left fingers showed marked thickening and distension of the flexor tendon sheath of the left index and middle fingers (Figure 2) with multiple fluid pockets within the tendon sheaths and areas of increase colour Doppler signal (Figure 3) in keeping with neovascularisation noted within the thickened synovium. On the right, ultrasound examination showed hypoechoic soft tissue swelling on the volar side of right middle finger distal phalanx lying adjacent

to the tendon insertion. It showed peripheral and internal flow on colour Doppler assessment (Figure 4).

Magnetic Resonance (MR) imaging of the left fingers showed marked tendon sheath thickening with increased fluid surrounding the left index and middle fingers flexor digitorum superficialis and flexor digitorum profundus tendons (Figure 5, 6 and 7). There was diffuse thickening of the subcutaneous soft tissue but no significant bone marrow oedema, bone destruction or joint involvement. On the right side, MR imaging showed soft tissue swelling of the DIP joint of the middle finger epicentred around the tip distally (Figure 8) with bone marrow oedema and cortical bone loss resulting in narrowing of the circumference of the mid shaft (Figure 9). Bone marrow oedema was also seen involving other phalanges.

Histological examination of an ultrasound targeted biopsy of the right middle finger lesion revealed inflammation with non-caseating granulomas in keeping with sarcoidosis. Further investigations including chest radiograph (Figure 10), lung function tests and electrocardiogram did not reveal any other organ involvement. Patient was started on oral steroids (prednisolone 20mg daily) and within 4 weeks there was a good response with a significant reduction in swelling and improved movement in all of the affected fingers. Methotrexate was added as a steroid sparing agent and the dose gradually increased to 25mg weekly whilst the steroids have been gradually reduced.

Patient remained static after two years of follow up. No new symptoms were reported on follow up but he did not notice improvement with methotrexate. Follow up MR examination of the left hand (Figure 12) approximately two years from initial examination did not show significant radiological changes with extensive tenosynovitis of the index and middle fingers flexor tendons. No tendon tear, bone marrow oedema or joint involvement was identified.

DISCUSSION

Etiology & Demographics

Sarcoidosis is an inflammatory disorder of unknown cause characterized by the presence of non-caseating granulomas in the tissues. The disease can affect multiple organs but most commonly lung, lymph nodes, skin and eyes [1]. Thoracic radiologic abnormalities are seen at some stage in approximately 90% of patients, with an estimated 20% developing chronic lung disease leading to pulmonary fibrosis [2]. Interestingly, our patient did not have any evidence of lung involvement on radiographs or lung function tests.

Skeletal involvement is relatively rare and has been reported in 1-13% of patients with sarcoidosis, with an estimated average of 5% [3]. These include inflammatory arthritis, periarticular soft tissue swelling, dactylitis, bone involvement, myopathy and tenosynovitis [4]. Tenosynovitis is commonly described in association with rheumatic diseases, infection and trauma, however it is a rare manifestation of

sarcoidosis and very rare to be the sole presenting feature. Literature review showed 22 reported cases of sarcoid tenosynovitis [1,5-16] however granulomatous tenosynovitis was the sole presenting manifestation of sarcoidosis in only 3 patients [1,9,12]. No specific gender or age predilection identified.

Clinical & Imaging findings

Sarcoid tenosynovitis is usually localized to the wrists, fingers or ankles. Patients usually present with pain, paraesthesia, soft tissue swelling and/or limited range of motion in the involved joint [9]. There is no specific pattern of involvement of the hand and wrist, although the extensor compartment is more frequently affected than the flexor compartment [13,17]. Examination usually reveals subcutaneous nodules or soft tissue swelling of the tendon sheaths [18].

Tenosynovitis is difficult to diagnose on plain film radiographs. Ultrasound usually shows synovial sheath thickening, increase fluid within the tendon sheath and hyperaemia on Doppler imaging. MR imaging will demonstrate increase fluid within the tendon sheath as high signal on T2 with low/intermediate T1 signal, with surrounding oedema and contrast enhancement.

Isolated sarcoid tenosynovitis does not have specific radiological findings differentiating it from other causes making diagnosis challenging. Associated sarcoidosis bone involvement with classic radiological features of cyst-like radiolucent areas, lacelike honeycomb appearance, or extensive bone erosion with pathologic fractures would be helpful in establishing the diagnosis [3].

Treatment & Prognosis

The key treatment strategy is with medical treatment and/or surgical treatment. The main stay of medical treatment is with corticosteroid therapy while surgical treatment usually consists of excision of the granulomatous tissue with tenosynovectomy [14,17,19]. Methotrexate can be used as a second-line treatment in steroid-refractory cases, as a steroid sparing treatment regime, or in case of steroid adverse effects. It can also be used as a first-line treatment mostly in combination with steroid. The recommended initial dose is 5-15 mg once a week orally [20]. Persistent and progressive sarcoid tenosynovitis despite medical and/or surgical treatment is reported in 18% of patients [9], therefore combination of both treatments are often used to reduce chance of disease recurrence [5,8,14,17,19]. Few reports mention spontaneous disease resolution [14].

Differential Diagnoses

The differential diagnosis of sarcoid granulomatous tenosynovitis includes: tuberculosis (TB), other mycobacterial infections, rheumatoid arthritis, psoriasis and brucellosis [8].

Sarcoid tenosynovitis is often very difficult to differentiate from TB and often biopsy is needed. TB tenosynovitis usually appears as cyst-like cavities associated with expansion of the underlying diaphysis and bone sequestration [21].

Rheumatoid arthritis can result in tenosynovitis, joints capsule and ligaments inflammatory destruction with fibrosis and scarring mimicking granulation tissue. Radiologically, this can be difficult to differentiate from sarcoid tenosynovitis however the characteristic symmetric involvement of small hand joints and the presence of erosive changes are often helpful [22].

Unlike rheumatoid arthritis, which has a polyarticular presentation, early psoriatic arthritis can occur in any of the small hand joints, mostly as monoarthritis or oligoarthritis. Isolated tenosynovitis, especially of the flexor tendon sheaths resulting in dactylitis, or sausage digits, is well known in psoriatic arthritis. Several fingers can be involved simultaneously (polydactylitis). The presence of enthesitis, multifocal bone marrow oedema, periostitis, and extracapsular enhancement accompanying articular or tendon sheath synovitis is almost diagnostic of psoriatic arthritis [23].

Brucellosis is a worldwide zoonosis with few reported cases of secondary dactylitis [24,25]. This can be difficult to differentiate from sarcoid dactylitis however brucellosis musculoskeletal involvement is often associated with sacroiliitis [25].

Sarcoid tenosynovitis should be differentiated from Darier-Roussy Sarcoidosis. This is an uncommon cutaneous manifestation of sarcoidosis characterised by subcutaneous granulomatous nodules and bilateral hilar adenopathy. It typically affects the legs and forearms but not the digits [18].

TEACHING POINT

Extensive tenosynovitis has several differential diagnoses and whilst it is a rare presentation of sarcoidosis, it should be considered in patients who do not demonstrate other systemic manifestations of sarcoidosis or the typical radiological sarcoid bone involvement.

REFERENCES

1. Fodor D, Poanta L, Rogojan L. Dactylitis and bone lesions at the onset of sarcoidosis: a case report. *Pol Arch Med Wewn* 2008 Dec;118(12):774-7. PMID 19202958
2. Criado E, Sánchez M, Ramírez J et al. Pulmonary Sarcoidosis: Typical and Atypical Manifestations at High-Resolution CT with Pathologic Correlation. *Radiographics*, 2010 Oct;30(6):1567-86. PMID 21071476.
3. Koyama T, Ueda H, Togashi K et al. Radiologic manifestations of sarcoidosis in various organs. *RadioGraphics*, 2004 Jan-Feb;24(1):87-104. PMID 14730039.
4. Torralba KD, Quismorio FP Jr. Sarcoid arthritis: a review of clinical features, pathology and therapy. *Sarcoidosis Vasc*

Diffuse Lung Diseases, 2003 Jun;20(2):95-103. PMID 12870718.

5. Burns DA, Sarkany I. Sarcoidosis with arthritis and tenosynovitis. *Clinical and Experimental Dermatology* 1978 Dec;3(4):439-42. PMID 737888.
6. Uddenfeldt P, Bjelle A, Olsson T. Musculo-skeletal symptoms in early sarcoidosis. Twenty-four newly diagnosed patients and a two-year follow-up. *Acta Medica Scandinavica*, 1983;214(4):279-84. PMID 6660034.
7. Curco N, Pagerols X, Vives P. Subcutaneous sarcoidosis with dactylitis. *Clinical and Experimental Dermatology*, 1995 Sep;20(5):434-35. PMID 8593727
8. Merle M, Bour C, Foucher G, Saint Laurent Y. Sarcoid tenosynovitis in the hand. A case report and literature review. *Journal of hand surgery (Edinburgh, Scotland)*, 1986 Jun;11(2):281-6. PMID 3734581.
9. Carlson JA, Wei-Lee H, Kutzner H, Jones DM, Tobin E. Sarcoidal granulomatous tenosynovitis of the hands occurring in an organ transplant patient. *Journal of Cutaneous Pathology*, 2007 Aug;34(8):658-64. PMID 17640239.
10. Kellner H, Spathling S, Herzer P. Ultrasound findings in Lofgren's syndrome: Is ankle swelling caused by arthritis, tenosynovitis or peri-arthritis? *Journal of Rheumatology*, 1992 Jan;19(1):38-41. PMID 1556697.
11. Kollert F, Hadiprasetya ML, Probst C et al. Clinical images: Multilocular tenosynovitis as a manifestation of sarcoidosis. *Arthritis and Rheumatism*, 2012 Jan;64(1):326. PMID 21971997
12. Fodor L, Bota IO, Fodor M and Ciuce C. Sarcoid flexor tenosynovitis as a single early manifestation of the disease. *Journal of Plastic, Reconstructive and Aesthetic Surgery*, 2012 Aug;65(8):217-9. PMID 22472050.
13. Wang HT, Sunil TM, Kleinert HE. Multiple unusual complications after extensive chronic sarcoid tenosynovitis of the hand: A case report. *Journal of Hand Surgery*, 2005 May;30(3):610-14. PMID 15925175.
14. Lambert L, Riemer EC, Judson MA. Rapid development of sarcoid tenosynovitis. *Journal of Clinical Rheumatology*, 2011 Jun;17(4):201-3. PMID 21617553.
15. Bleton R, Alnot JY, Kahn MF, Bocquet L. Sarcoid synovitis. A case report of localization at the level of the flexor tendons of the fingers. *Ann Chir Main Memb Super*, 1991;10(4):360-3. PMID 1720975.
16. El Hassani S, Allali F, Lazrak N, Hajjaj-Hassouni N. Sarcoid tenosynovitis. Report of a case. *Rev Rhum Engl Ed* 1999 May;66(5):296-7. PMID 10380265.

17. Iannuzzi MC, Fontana JR. Sarcoidosis: clinical presentation, immunopathogenesis, and therapeutics. *JAMA* 2011 Jan;305(4):391-9. PMID 21266686.
18. Morganroth P, Lim H, Chaffins M. Sarcoidosis presenting as subcutaneous finger and toe nodules. *Journal of the American Academy of Dermatology*, 2013 Apr;68(4):supplement1:AB141.
19. Gonzalez del Pino J, Diez Ulloa A, Lovic A, Relea MF. Sarcoidosis of the hand and wrist: a report of two cases. *Journal of Hand Surgery*, 1997 Sep;22(5):942-5. PMID 9330159.
20. Cremersa J, Drenta M, Bastb A et al. Current opinion review. Multinational evidence-based World Association of Sarcoidosis and other Granulomatous Disorders recommendations for the use of methotrexate in sarcoidosis: integrating systematic literature research and expert opinion of sarcoidologists worldwide. 1070-5287 © 2013 Wolters Kluwer Health | Lippincott Williams & Wilkins.
21. De Backer AI, Vanhoenacker FM, Sanghvi DA. Imaging features of extraaxial musculoskeletal tuberculosis. *Indian Journal of Radiology and Imaging*, 2009 Jul-Sep;19(3):176-86. PMID 19881081.
22. Sommer OJ, Kladosek A, Weiler V et al. Rheumatoid arthritis: a practical guide to state-of-the-art imaging, image interpretation, and clinical implications. *Radiographics*, 2005 Mar-Apr;25(2):381-98. PMID 15798057.
23. Spira D, Kotter I, Henes J et al. MRI findings in psoriatic arthritis of the hands. *AJR*. 2010 Nov;195(5):1187-93. PMID 20966327.
24. Ozgocmen S, Ardicoglu O, Ozcakar L. Dactylitis in a patient with brucellosis. *Journal of hand surgery Br*, 2001 Apr;26(2):171-2. PMID 11281675.
25. Batmaza I, Tekinb R, Sariyildiza MA et al. A Case of Brucellosis With Simultaneous Dactylitis and Sacroiliitis. *Journal of Medical Cases*, 2012;3(5):304-307.

FIGURES

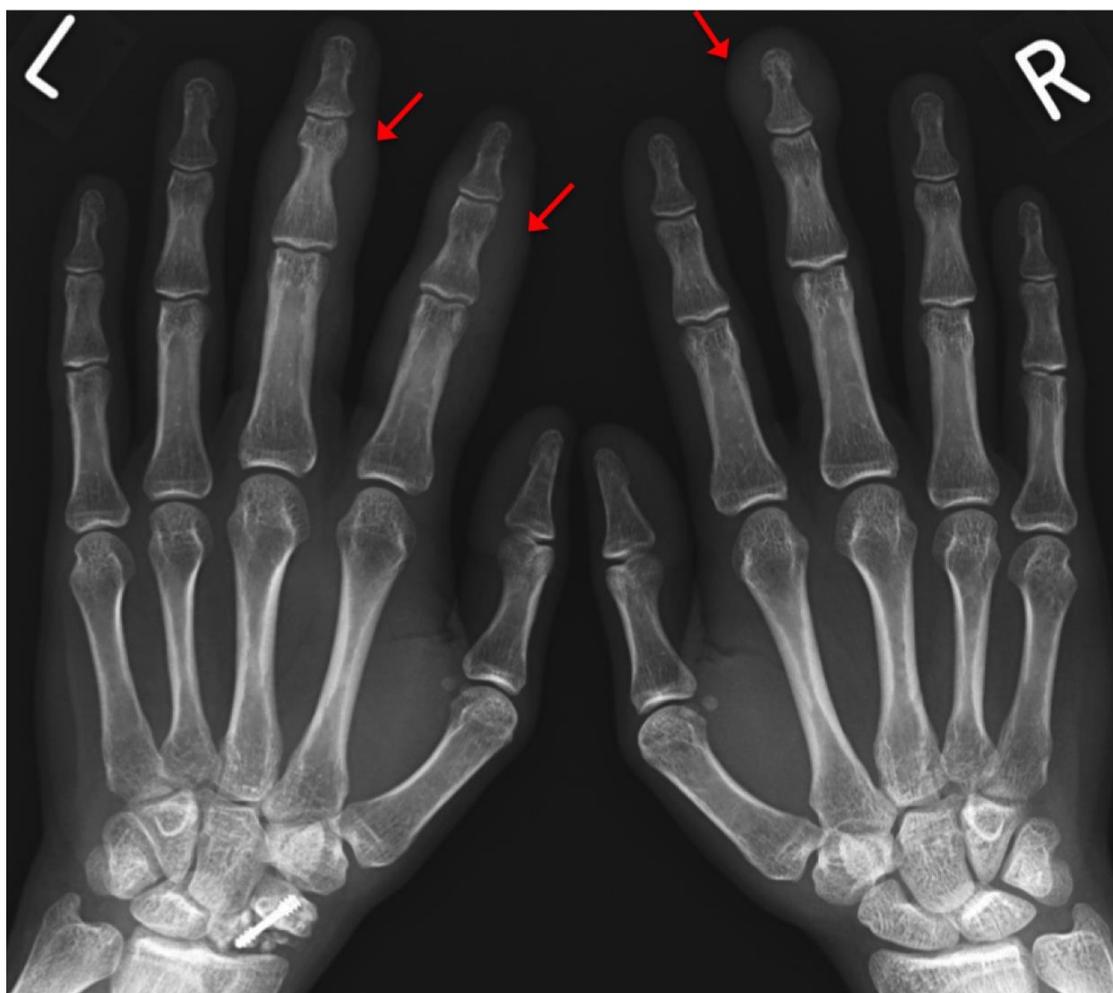


Figure 1: A 42 year old male with sarcoid tenosynovitis. Bilateral hand radiograph shows fusiform soft tissue swelling over left index and middle fingers phalanges and right middle finger distal phalanx (arrows). No bone involvement, cortical destruction or arthropathy demonstrated.

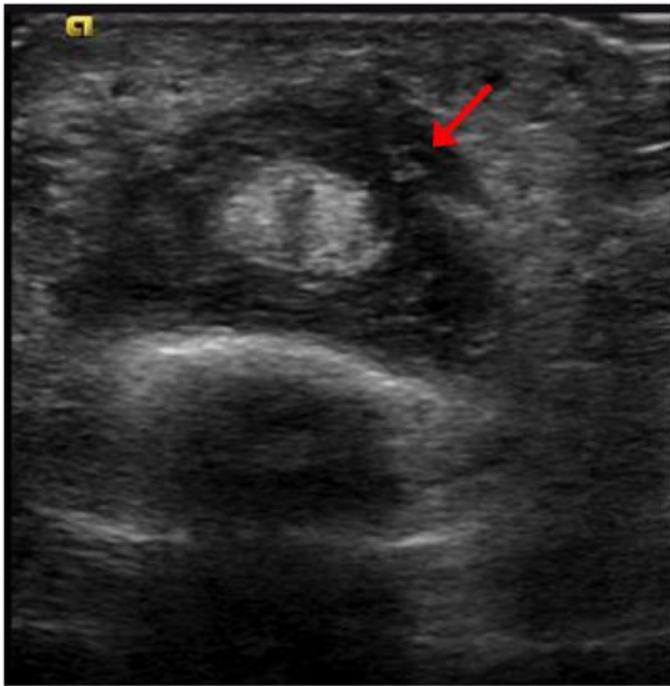


Figure 2: A 42 year old male with sarcoid tenosynovitis. Ultrasound examination of the left index finger in transverse plane showing extensive thickening of the flexor sheath (arrow). Technique: ultrasound examination using high frequency (12 Megahertz) linear probe with superficial musculoskeletal settings.

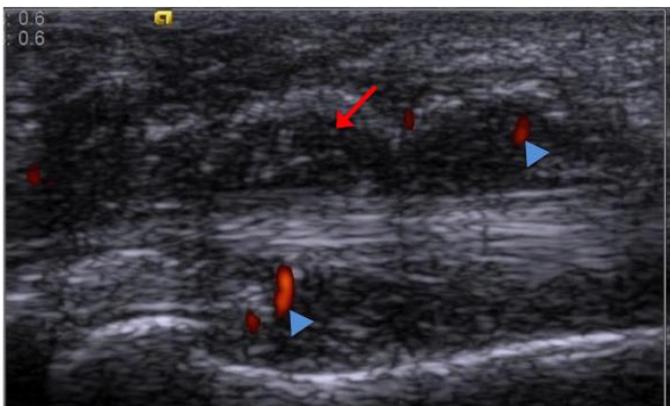


Figure 3: A 42 year old male with sarcoid tenosynovitis. Ultrasound examination of the left middle finger over the PIP joint in longitudinal plane demonstrates flexor tendon thickening (arrow) with increase vascularity on colour Doppler study (arrow heads). Technique: ultrasound examination using high frequency (12 Megahertz) linear probe with superficial musculoskeletal settings.

Figure 5 (right): A 42 year old male with sarcoid tenosynovitis. Sagittal T2 weighted MR image through the left middle finger showing fluid surrounding the flexor digitorum superficialis and flexor digitorum profundus tendons (arrows) with significant superficial soft tissue swelling (arrow head). Technique: MR Imaging; magnet strength 1.5T. TE = 82 msec and TR = 3950 msec. Small field of view with surface coil. No contrast.

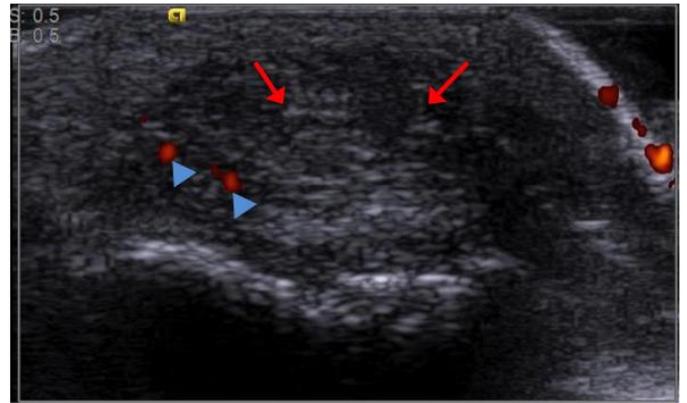


Figure 4: A 42 year old male with sarcoid tenosynovitis. Ultrasound examination over the distal phalanx of the right middle finger in longitudinal plane demonstrates large soft tissue swelling (arrows) measuring 3 x 2.5 x 2 cm, overlying the distal phalanx with homogenous internal echotexture and increase vascularity (arrow heads). Technique: ultrasound examination using high frequency (12 Megahertz) linear probe with superficial musculoskeletal settings.



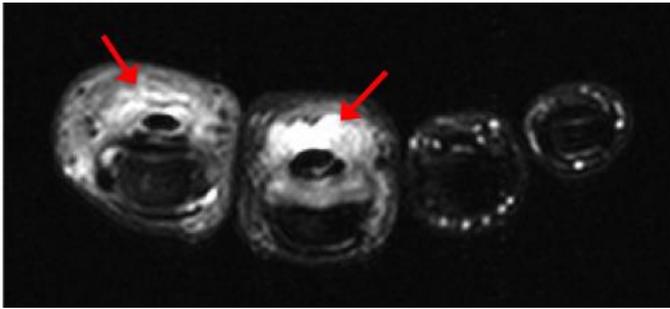


Figure 6: A 42 year old male with sarcoid tenosynovitis. Axial fat saturated T2 weighted MR image through the left index and middle fingers showing marked tendon sheath thickening with increased fluid surrounding the flexor digitorum superficialis and flexor digitorum profundus tendons (arrows). Technique: MR Imaging; magnet strength 1.5T. TE = 82 msec and TR = 5300 msec. Small field of view with surface coil. No contrast.

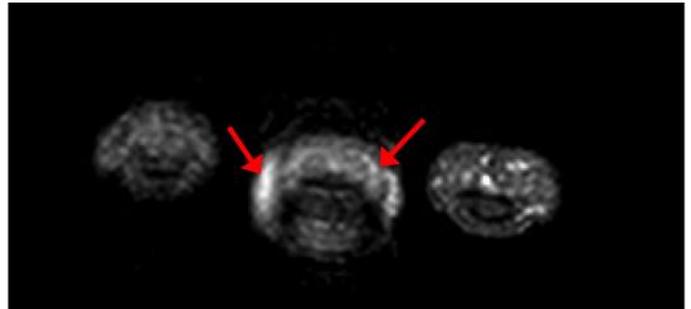


Figure 8: A 42 year old male with sarcoid tenosynovitis. Axial fat saturated T2 weighted MR image through the right middle finger showing soft tissue swelling over the volar aspect of the distal phalanx (arrows). Technique: MR Imaging; magnet strength 1.5T. TE = 82 msec and TR = 5830 msec. Small field of view with surface coil. No contrast.



Figure 7: A 42 year old male with sarcoid tenosynovitis. Coronal T1 turbo inversion recovery magnitude (TRIM) sequence MR image through the left index and middle fingers showing marked tendon sheath thickening with increased fluid surrounding the flexor digitorum superficialis and flexor digitorum profundus tendons (arrows) with significant superficial soft tissue swelling (arrow heads). Technique: MR Imaging; magnet strength 1.5T. TE = 29 msec and TR = 4000 msec. Small field of view with surface coil. No contrast.



Figure 9: A 42 year old male with sarcoid tenosynovitis. Coronal T1 weighted MR image through the right middle finger showing marrow oedema (arrow head) and cortical bone loss (arrow) resulting in narrowing of the circumference of the mid shaft. Technique: MR Imaging; magnet strength 1.5T. TE = 21 msec and TR = 847 msec. Small field of view with surface coil. No contrast.

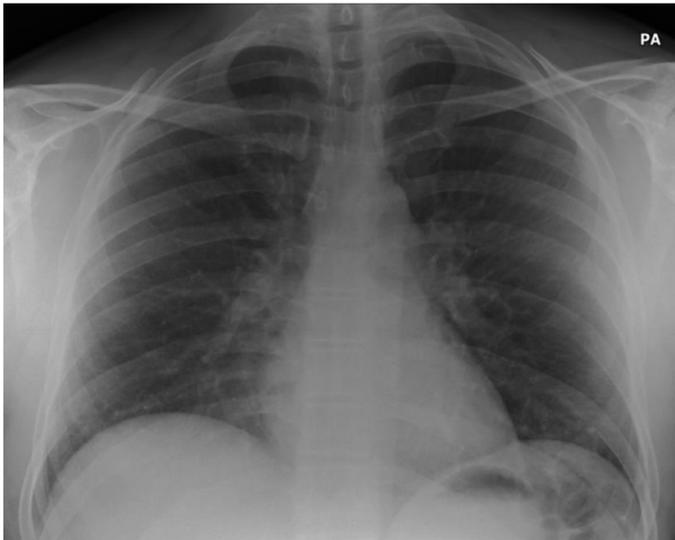


Figure 10: A 42 year old male with sarcoid tenosynovitis. Chest radiograph shows clear lungs with no evidence of interstitial changes, hilar or mediastinal lymphadenopathy.



Figure 12: A 42 year old male with sarcoid tenosynovitis. Follow up MR examination approximately 2 years from initial examination. Coronal fat saturated T2 weighted MR image through the left index and middle fingers showing extensive thickening of the index and middle fingers flexor tendons with surrounding lobulated, well defined, cystic areas within the tendon sheaths (arrows) showing high T2 signal. Appearance is in keeping with tenosynovitis of the index and middle fingers flexor tendons. Technique: MR Imaging; magnet strength 1.5T. TE = 24 msec and TR = 1830 msec. Small field of view with surface coil. No contrast.

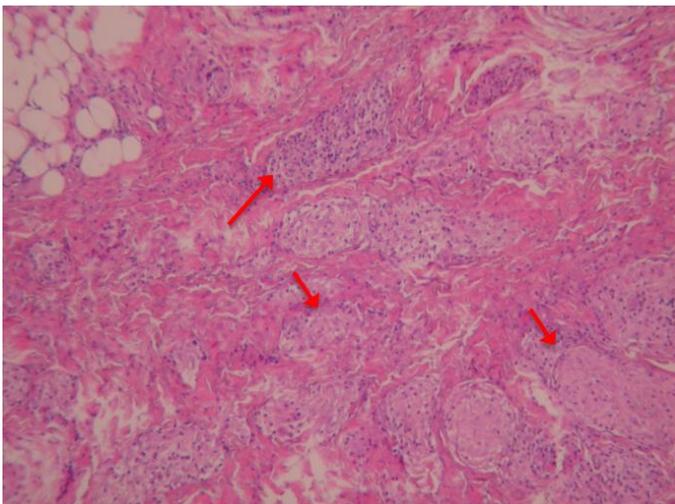


Figure 11: A 42 year old male with sarcoid tenosynovitis. Microscopic slide specimen of tissue biopsy obtained from right middle finger distal phalanx soft tissue mass (Hematoxylin and eosin stain, 20x), showing extensive non-caseating granulomas (arrows) in keeping with sarcoid disease.

Aetiology	Idiopathic inflammatory disorder
Incidence	Very rare (few case reports and series published)
Gender ratio	None
Age predilection	None
Risk factors	None
Treatment	Steroids, immunomodulation therapy.
Prognosis	Persistent and progressive disease reported in 18% of cases
Findings on imaging	Plain radiograph shows soft tissue swelling +/- classic radiological features of cyst-like radiolucent areas. Ultrasound shows tendon sheath thickening, oedema and hyperaemia. MRI demonstrates oedema signal changes (low signal on T1, high signal on T2) with thickening and enhancement of the involved tendon sheaths. These changes may extend to the adjacent bone and periarticular soft tissue.

Table 1: Summary table of sarcoid tenosynovitis.

	Plain radiograph	Ultrasound	MRI
Sarcoid tenosynovitis	Soft tissue swelling +/- classic radiological features of cyst-like radiolucent areas.	Tendon sheath thickening, oedema and hyperaemia.	Oedema signal changes (low signal on T1, high signal on T2) with thickening and enhancement of the involved tendon sheaths. These changes may extend to the adjacent bone and periarticular soft tissue.
TB tenosynovitis	Soft tissue swelling +/- bone involvement (osseous erosions and sequestration).	Thickening/granuloma of the involved tendon with increase colour Doppler signal.	Cyst-like lesions (high signal on T2) with diaphysis expansion and bone sequestration. Tendon sheath enhancement is present.
Rheumatoid arthritis	Soft tissue swelling, characteristic Rheumatoid arthritis arthropathy.	Thickening, hyperaemia and oedema of tendon sheath. Associated synovitis and erosive arthropathy of small joints of hand.	High signal around the tendon sheath on T2 in case of tenosynovitis. Low signal lesions on T1 and T2 in case of fibrosis/scarring. Presences of periarticular erosions and small joints inflammatory changes. Tendon sheath enhancement is present.
Psoriasis	Soft tissue swelling (sausage fingers), characteristic psoriatic arthropathy changes.	Mainly flexor tendon sheaths thickening, oedema, hyperaemia and associated enthesitis.	High T2 signal and thickening of flexor tendons sheath with extension of signal changes into tendon insertion (enthesitis) and multifocal bone marrow oedema, and periostitis. Enhancement is present.
Brucellosis	Soft tissue swelling +/- sacroiliitis.	Tendon sheath oedema and thickening.	High T2 signal changes within the involved tendon sheath. High T2 signal/fluid within sacroiliac joints. Tendon sheath enhancement is often present.

Table 2: Differential table for sarcoid tenosynovitis.

ABBREVIATIONS

DIP = distal interphalangeal
PIP = proximal interphalangeal
MR = Magnetic Resonance
TB = tuberculosis
TRIM = Turbo Inversion Recovery Magnitude

KEYWORDS

Sarcoidosis; tenosynovitis; hand; phalanges; Magnetic Resonance Imaging; MRI

Online access

This publication is online available at:
www.radiologycases.com/index.php/radiologycases/article/view/2311

Peer discussion

Discuss this manuscript in our protected discussion forum at:
www.radiolopolis.com/forums/JRCR

Interactivity

This publication is available as an interactive article with scroll, window/level, magnify and more features.
Available online at www.RadiologyCases.com

Published by EduRad



www.EduRad.org