

Drumstick Dactylitis: An Unusual Presentation Of Sarcoid

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Citation

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Abstract

We report a case of a 34-year-old African American man with bulbous sarcoidosis of the digits, which we term drumstick dactylitis. The patient presented with bulbous swelling of the distal finger and toe with associated nail dystrophy. This is the third reported case of bulbous sarcoidosis in the English-language literature.

INTRODUCTION

Sarcoidosis is a multisystem disease of unknown etiology that involves predominantly the lungs, lymph nodes, skin, and eyes.¹ It has been noted to occur more commonly in African Americans.¹ Cutaneous involvement occurs in as many as one third of patients with sarcoidosis.¹ Multiple cutaneous morphologies of sarcoidosis are possible, some of which are quite rare. Sarcoidosis of the finger is one such presentation. We describe a case of a 34-year-old African American male with bulbous sarcoidosis of the digits. To our knowledge, this cutaneous manifestation has only been reported twice before.

CASE REPORT

A 34-year-old black man was admitted for dyspnea and pleuritic chest pain and was referred to Dermatology for evaluation for possible onychomycosis. He had a history of 3rd degree A-V heart block and suspected pulmonary tuberculosis five years earlier for which he was treated despite negative acid-fast bacilli smears, tuberculin skin tests, and culture of pleural fluid. Pleural biopsy at that time showed ill-formed non-necrotizing granulomas.

On current examination the patient had a violaceous bulbous swelling at the distal end of the left 4th finger with almost vesicular appearing red macules on the tip. The left 4th fingernail was dystrophic, yellowed, and thickened with pronounced longitudinal ridging. The right 2nd toe was similarly bulbous with nail dystrophy, and the right great toe had a red-purple firm plaque in the hyponychium. The left thumbnail had central longitudinal ridging and a purple patch overlying the matrix region. The other nails appeared normal and there were no other cutaneous findings.

Differential diagnosis included tuberculosis, deep fungal infection, sarcoidosis, and metastatic malignancy.

A 4mm punch biopsy was taken from the tip of left 4th finger. The biopsy specimen showed non-necrotizing epithelioid granulomas. No birefringent foreign particles were found on polarization microscopy. The Fite and periodic acid-Schiff stains were negative for acid-fast mycobacteria and fungal organisms, respectively. Tissue cultures were negative for bacteria, fungi, and mycobacteria.

Radiograph of the hand showed a bony erosion of the left 4th distal phalanx. The patient also had interstitial lung infiltrates, bilateral hilar adenopathy, left pleural effusion, and right paratracheal mass on high resolution computerized axial tomography scan of the chest. Further investigations included normal serum calcium of 8.6 mg/dL (normal range, 8.6mg/dL-10.6mg/dL), normal liver function tests, slightly abnormal serum angiotensin-converting enzyme level of 68 IU/L (normal range, 9-67 IU/L) and non-reactive tuberculin skin test. Cultures of pleural fluid aspiration were negative for acid-fast bacilli and fungus. Sputum smears and cultures for acid-fast bacilli were negative. Serologic tests for HIV and syphilis were negative. Coccidioides and Histoplasma antibodies were negative.

A diagnosis of systemic sarcoidosis with dactylitis was made. The patient was subsequently started on oral prednisone, 40mg/day because of lung and bone involvement. After three weeks, significant improvement was noted in the lesion of left 4th finger with a decrease in both the violaceous discoloration and swelling. Intralesional triamcinolone was started at that time.

DISCUSSION

Finger involvement in patients with sarcoidosis is well recognized. Manifestations of the finger include multiple nodules,² pseudoclubbing, true clubbing,³ subcutaneous nodules with ulceration,⁴ finger pain without soft tissue changes,⁵ and dactylitis.

Sarcoid dactylitis implies involvement of bone and soft tissue of the fingers, most classically presenting with bilateral^{6,7,8} fusiform or sausage-shaped swellings.⁸ Dactylitis is a rare manifestation occurring in only 0.2% of patients with sarcoidosis,^{9,10} and is often associated with lupus pernio.¹¹ In our patient, bony erosion of the distal phalanx was evident on radiograph, although he did not present with the classic fusiform swelling of the soft tissue. Our patient presented with an unusual bulbous swelling of the digits which we term “drumstick dactylitis”.

In our review of the English-language literature, we could find only two reports of patients with a similar “drumstick” shaped dactylitis. In the first case, Pitt et al,⁹ describe a 50-year-old West Indian woman with bulbous involvement of her right 4th and left 3rd toes. The bulbous swellings in this case contrast ours in that they were bilateral and occurred more proximally on the affected digit instead of on the fingertips. Jacyk⁸, in 1999, described eight South African patients exhibiting sarcoid dactylitis with nail changes similar to our patient. Of the eight patients reported, five had lupus pernio, two had a mutilating form, and one had bone changes.⁸ In contrast to the unilateral finger involvement in our patient, all eight patients in this report were found to have bilateral involvement.⁸ A slightly different presentation reported by Di Landro et al,² describes a patient with multiple large nodules localized to the tips of several fingers without associated nail involvement.

Nail involvement in sarcoidosis is rare.^{12,13} However, in patients with sarcoid dactylitis, nail involvement occurs much more frequently.^{6,9} Nail involvement may manifest as distal onycholysis,¹⁴ subungual hyperkeratosis,¹⁵ dystrophy,¹³ true clubbing,¹² and pterygium formation.¹⁶ Infiltration of the nail and nearby skin may cause fissuring and swelling.¹³ Other nail changes include longitudinal ridging, cracking, thickening, brittleness, fragility and pitting.¹² Nail dystrophy is associated with a chronic disease course and underlying bone involvement.¹² Due to this association, sarcoidosis

accompanied by nail dystrophy warrants radiological examination of the hands and feet. In addition, Mann et al¹³ suggest a nail biopsy in patients with nail dystrophy of unknown cause to assess for sarcoidosis.

In conclusion, we describe a bulbous swelling of the distal finger and toe with associated nail dystrophy, which differs from classic sarcoid dactylitis. Drumstick dactylitis is the suggested term for this rare form of sarcoid.

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