Case report

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Rice bodies, extensor digitorum longus tendon tenosynovitis and sarcoidosis – multimodal ultrasound approach. A case report.

Daniela Fodor[1], Maria Bădărînză[1], Claudiu Filip[2], Doinița Crișan[3], Oana Șerban[1]

[1]2nd Internal Medicine Department, "Iuliu Haţieganu" University of Medicine and Pharmacy, [2]Plastic Surgery Department, Emergency County Hospital, [3]Pathology Department, Emergency County Hospital, Cluj-Napoca, Romania

Abstract

We present the case of a young woman in whom the association between sarcoidosis, extensor digitorum longus tendon tenosynovitis, and the presence of numerous rice bodies within the tendon sheath revealed a new clinical manifestation of sarcoidosis, in the absence of pulmonary or lymph nodes involvement. The most important aspect of this case is the assessment of tenosynovitis using multimodal ultrasound (grey scale, color/power Doppler, strain elastography, contrast enhanced ultrasound) which demonstrated specific findings that may facilitate further recognition of this clinical condition.

Keywords: rice body; sarcoidosis; tenosynovitis; ultrasound

Introduction

Rice bodies, first described by Riese in 1895 as small particles resembling grains of rice, may be found in synovial spaces, including joints, bursae, tendon sheaths. They have been associated with mycobacterial infections, rheumatoid arthritis, systemic lupus erythematosus [1-3], and other conditions; however, in some cases, no etiology can be demonstrated [4]. Sarcoidosis is an inflammatory granulomatous systemic disease that may affect any organ or system, although the lungs and lymph nodes are most frequently involved [5,6]. Musculoskeletal manifestations of sarcoidosis occur in one-third of patients and include Löfgren syndrome (acute arthritis associated with hilar lymphadenopathy and erythema nodosum), chronic arthritis, Jaccoud arthropathy, spinal involvement, dactylitis, and myopathy [5,7]. Tenosynovitis has been described in the extensor compartments of the wrist and, rarely, at the ankle. To the best of our knowledge, no case describing the association between rice bodies, ankle tendons tenosynovitis, and sarcoidosis has been published, and we found no description of rice bodies assessed by multiparametric ultrasound (US).

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Corresponding author: Daniela Fodor, MD, PhD
2nd Internal Medicine Department
"Iuliu Haţieganu" University of Medicine and Pharmacy, Cluj-Napoca, Romania
2-4 Clinicilor str, 400006 Cluj-Napoca, Romania
E-mail: dfodor@umfcluj.ro

Case report

A previously healthy 27-year-old woman presented with tender, erythematous subcutaneous nodules in the pretibial areas and pain in the left ankle. As the clinical appearance was highly suggestive for erythema nodosum, a systematic work-up was initiated to identify the underlying etiology. Except for low grade fever, no other general symptoms were identified. Sarcoidoisis was initially suspected, in the context of possible Löfgren's syndrome, but the thoracic computed tomography scan was normal. Serum angiotensin-converting enzyme (ACE)

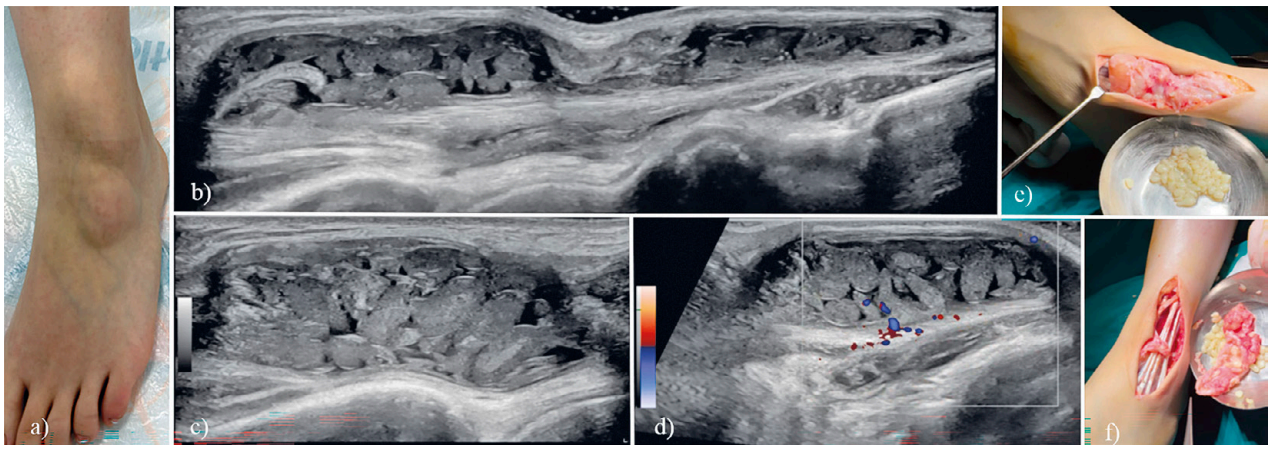

Fig 1. a) Clinical aspect of the extensor digitorum longus tendon tenosynovitis; b) and c) panoramic longitudinal/transversal scan of the extensor digitorum longus tendon; note the distended tendon sheath fulfilled with round/oval regular nodules, hypoechoic, some with central hyperechogenicity; d) no color was obtained at color Doppler examination: e) intraoperative aspect of tenosynovitis and rice bodies extracted from tendon sheath; f) the aspect of the "cleaned" tendon at the end of the surgical procedure.

was slightly increased (64U/L compared with a normal upper limit of 52U/L), and US detected a small amount of fluid in the left ankle. Other possible etiologies were investigated (tuberculosis, streptococcal infections, Behçet disease, inflammatory bowel disease, drug related causes, etc.), but no diagnosis could be confirmed. The clinical course was favorable after 10 days of treatment with naproxen, 500 mg twice daily.

A few months later, the patient returned with low-grade pain in the left ankle and progressive swelling in the anterolateral ankle region. As ankle effusion and extensor digitorum longus tendon tenosynovitis were identified by US, local corticosteroid injection into the tendon sheath was administered, with no effect. After another two months, marked tenosynovitis of the common extensor digitorum longus tendons sheath was evident on clinical examination (fig 1a), and the patient underwent repeat US examination (LOGIQ™ E10 Ultrasound/GE Healthcare, United States). On gray-scale US, the tendon sheath was distended and filled with solid material organized into nodules. The nodules were hypoechoic, most of them with a hyperechoic center, and all had regular margins (fig 1 b,c, video 1 and 2 on the journal site). The appearance was initially interpreted as synovial hypertrophy; however, no vascularization was detected on Doppler examination (fig 1d), and information regarding nodules stiffness could not be obtained by strain elastography, as adequate color filling of the region of interest could not be achieved. For these reasons, contrast-enhanced US (CEUS) was performed using 2.4 ml SonoVue, but no contrast agent entered these nodules (video 3, on the journal site), confirming that they were not vascularized and did not represent synovial pannus or other type of proliferative parenchymal tissue. Therefore, the suspicion of rice bodies was raised. The patient was referend to the plastic surgery department. Intraoperatively, rice bodies surrounded by granulomatous tissue were found in the extensor tendon sheath (fig 1 e,f). Histopathological examination revealed multiple non-caseating granulomas. In a few granulomas, a small central area of fibrinoid necrosis was identified, but periodic acid–Schiff (PAS) and Ziehl-Neelsen staining were negative. The case was finally interpreted as sarcoid tenosynovitis associated with rice bodies. Postoperatively, no systemic treatment was considered necessary, but the patient remains under regular follow-up.

Discussion

We report this case for two reasons. First, to the best of our knowledge, the association between rice body, sarcoidosis and extensor digitorum longus tendon involvement was not been previously reported. Second, we aimed to describe the appearance of rice bodies using multimodal US examination.

The diagnosis was challenging because the initial clinical picture was only partially suggestive of sarcoidosis. The association between erythema nodous and sarcoidosis is well known, especially in acute presentations [5,7,8]. Demonstration of non-necrotizing epithelioid cell granulomas is important establishing the diagnosis; however, necrosis within sarcoid granulomas has also been reported in approximately 3–30% of cases [9]. In our case two of the classic criteria for Löfgren syndrome [5-8], namely erythema nodosum and ankle involvement, were present, but the absence of mediastinal lymphadenopathy delayed the diagnosis, even if ACE level was increased.

Musculoskeletal involvement in sarcoidosis is heterogeneous. Although ankle involvement is frequent in acute presentations, true ankle arthritis, with US-demonstrated synovitis and Doppler activity, is in fact rare; soft tissue swelling (edema) or tenosynovitis (with Doppler signal in many cases) is encountered more frequently [5,7,10]. The present case reinforces this concept, as the dominant abnormality was a marked tenosynovitis of the extensor digitorum longus tendon, rather than joint synovitis. The presence of rice bodies within the tendon sheath further broadened the differential diagnosis.

Rice bodies are nonspecific structures that may occur in chronic inflammatory or infectious synovial diseases. They are amorphous, acidophilic, cartilage-like bodies, composed of an outer fibrin layer and a collagen core, but the exact mechanism of their formation remains a matter of debate [1-4]. Therefore, identification of rice bodies alone does not establish the underlying disease. In the present case, their association with granulomatous tenosynovitis and the clinical context of erythema nodosum favored sarcoidosis as the most plausible etiology. Most published data consist of case reports, and no published study has analyzed their specific imaging appearance. Cheung et al [11] analyzed published magnetic resonance imaging (MRI) reports in 22 patients and concluded that rice body-like synovial particles identified on MRI corresponded histologically to true rice bodies in only 45% of cases.

There are few reports describing rice bodies using US [3,12-14], and we found no data regarding their multiparametric US appearance. On gray-scale US, rice bodies are round or oval, well-defined, and hypoechoic, sometimes with a hyperechoic center. They show no vascularization on Doppler examination or CEUS, and their stiffness cannot be demonstrated using strain elastography. Tian et al [3] suggest that US has limited utility in the diagnosis rice bodies, as only 3 of the 23 patients reported in the published literature were diagnosed by US. This situation may be explained by the particular US appearance of rice bodies, the absence of published identification criteria, and probably the limited experience of examiners in recognizing them.

In conclusion, sarcoidosis, known as “the great imitator”, has a wide variety of extrapulmonary clinical manifestations. In the presence of rice bodies, sarcoidosis should be considered in the differential diagnosis. The US appearance of rice bodies is typical: they are hypoechoic, round, well delineated, show no vascularization with any US techniques, and cannot be characterized using strain elastography.

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Video legends

Video 1. Longitudinal scan of the extensor digitorum longus tendon

Video 2. Transvers scan of the extensor digitorum longus tendon

Video 3. CEUS examination – no contrast agent was uptake by the solid nodules of distended tendon sheath during the first minute of examination.