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Single Case

A Case of Subcutaneous Sarcoidosis Occurring along the Superficial Veins of the Forearms: A Distinctive Cutaneous Manifestation Masquerading Venous Tropic Action in the Underlying Systemic Disease?

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Abstract
Sarcoidosis is a multisystem disease of unknown etiology, developing granulomas in any tissues and organs. Approximately 25% of sarcoidosis patients have cutaneous involvement with various clinical manifestations, which are categorized into specific or nonspecific diseases based on the histopathology; the former represents the typical sarcoid granulomas. Subcutaneous sarcoidosis is one of the specific skin lesions and often affects extremities, to a much lesser extent with other anatomical sites. Herein, we report the case of an 82-year-old Japanese man with subcutaneous sarcoidosis whose skin nodules exclusively overlay the lines of superficial veins on the forearms. This rare clinical presentation was discussed with the literature reported thus far to access the underlying disease pathophysiology from the viewpoint of tropic response to the venous system in systemic sarcoidosis.

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Case Report

An 82-year-old Japanese man complained of a 1-month history of otherwise asymptomatic papules and nodules distributing on the abdomen, shoulders, and extremities. Some of the abdominal skin lesions corresponded with the sites of subcutaneous insulin injection therapy for uncontrollable type 2 diabetes mellitus. Of note, the skin nodules clustering on the forearms tended to overlie exclusively the median basilica, cubital, or cephalic veins (Fig. 1a, b). These lesions showed fluctuation during his clinical course, irrespective of the skin sites of periodic blood sampling and insulin injection, and further extended along the peripheral venous lines of his forearm skin (Fig. 1c, d). Transcutaneous ultrasonography of the forearm lesions revealed demarcated, homogeneous, hypoechoic areas surrounding superficial veins, suggestive of peri-/intravascular granuloma (Fig. 1e, f). Laboratory tests showed elevated levels of ESR (39 mm/h), CRP (1.07 mg/dL), sIL-2R (1,803 U/mL), and HbA1c (7.3%), without increase in serum calcium and ACE levels. The QuantiFERON-TB and tuberculin skin tests were negative. Owing to the refusal to obtain the skin biopsy from his forearm lesions, the skin sampling was undertaken from the abdominal lesion. The histopathology showed a nonnecrotic, noncaseating epithelioid granuloma with a relatively lower infiltration of inflammatory cells in the lower dermis and adipose tissue (Fig. 2). Bilateral hilar lymphadenopathy (BHL) and a solitary mass in the left gluteus minimus muscle were detected on positron emission tomography screening (Fig. 3a, b, respectively). Ophthalmological and cardiovascular examinations did not show any granulomatous lesions. Based on these findings, we made a diagnosis of sarcoidosis involving the skin, skeletal muscle, and BHL. During 1 year of follow-up, his abdominal skin lesion almost diminished with topical steroid therapy (Fig. 4a), but the forearm skin lesions often recurred along the venous lines (Fig. 4b, c). His blood test revealed improvement of the sIL-2R level (677 U/mL) but BHL remained unchanged.

Discussion

Only 7 reports, including our present case, have described sarcoidosis in which cutaneous involvement occurred along the superficial veins [1–6]. Clinically, their skin lesions were manifested by papules or nodules. Of these 7 cases, 5 (83.3%) had long been treated with intravenous injection of interferon-α for chronic hepatitis C, and their sarcoid skin lesions were related to the exact injected sites [1–5], implicating koebnerization and/or interferon-induced disease activity. Four cases (66.7%) also developed other organ involvement with a high incidence of BHL. Therefore, apart from the local interferon therapy, our case is the first report of de novo sarcoidosis demonstrating venous tropic skin lesions.

Little is known about the potential significance of the venous tropic manifestation in cutaneous sarcoidosis, but evidence may implicate the relatively pronounced trend of the disease pathology towards the systemic venous unit; for example, a retrospective study (n = 32) demonstrated that one-third (30.8%) of cutaneous sarcoidosis lesions microscopically had vascular involvement, ~70% of which (8/12, 66.7%) affected the venous plexuses in the dermal vasculature [5]. More specifically, a pathological examination of 40 autopsied sarcoid cases revealed a 2.4-times higher incidence of venous involvement in their pulmonary granulomas compared to the arterial involvement in the same organ [6]. An extrapolation of these observations and our present case may thus encompass the plausible interpretation that the venous tropic action in cutaneous sarcoidosis is more likely to be the pathogenic

counterpart of systemic venous involvement, like a clinically occult skeletal muscle lesion in our case (Fig. 3b). Since the vast majority of cutaneous sarcoidosis cases are asymptomatic, the venous tropic skin manifestation may be an underrecognized entity, albeit having a much higher occurrence in the entire disease spectrum. Dermatologists need to be aware of this otherwise unnoticeable skin manifestation as a distinct cutaneous phenotype masquerading systemic microangiopathy in sarcoidosis.

Statement of Ethics

The patient has given his informed consent for the publication of his case.

Disclosure Statement

The authors have no conflicts of interest to disclose. There were no funding sources for this work.

References

1 Shuja F, Kavoussi SC, Mir MR, Jogi RP, Rosen T: Interferon induced sarcoidosis with cutaneous involvement along lines of venous drainage in a former intravenous drug user. Dermatol Online J 2009;15:4.
2 Lee YB, Lee JI, Park HJ, Cho BK, Oh ST: Interferon-alpha induced sarcoidosis with cutaneous involvement along the lines of venous drainage. Ann Dermatol 2011;23:239–241.
3 Doyle ML, Berggren R, Magnus JH: Interferon-induced sarcoidosis. J Clin Rheumatol 2006;12:241–248.
4 Buss G, Cattin V, Spring P, Malinverni R, Gilliet M: Two cases of interferon-alpha-induced sarcoidosis koebnerized along venous drainage lines: new pathogenic insights and review of the literature of interferon-induced sarcoidosis. Dermatology 2013;226:289–297.
5 Takemura T, Shishiba T, Akiyama O, Oritsu M, Matsui Y, Eishi Y: Vascular involvement in cutaneous sarcoidosis. Pathol Int 1997;47:84–89.
6 Takemura T, Matsui Y, Saiki S, Mikami R: Pulmonary vascular involvement in sarcoidosis: a report of 40 autopsy cases. Hum Pathol 1992;23:1216–1223.

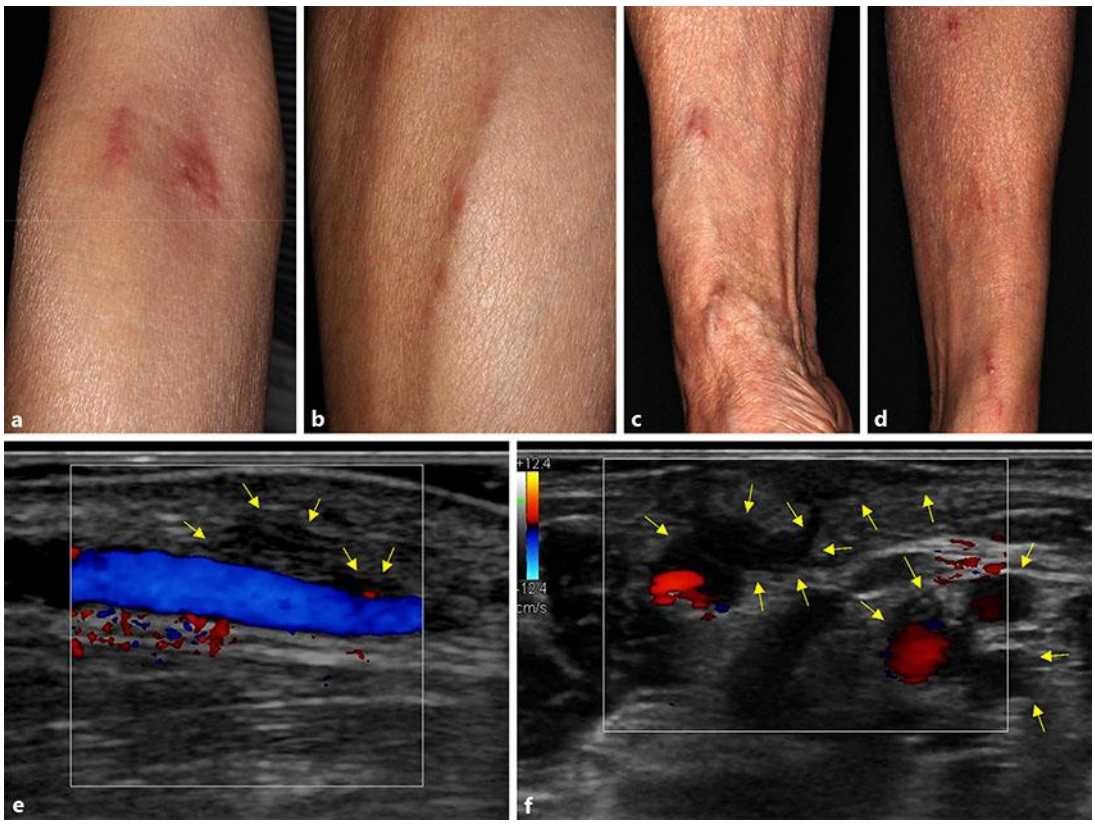

Fig. 1. Multiple papules and nodules along with the superficial venous lines on the flexor (**a**) and extensor aspects of the right forearm (**b**). The newly developed lesions along the peripheral cephalic veins of the right (**c**) and (**d**) left forearms. Transcutaneous ultrasonography showing hypoechoic masses that surround the superficial vein, as indicated by yellow arrows (**e**, **f**).

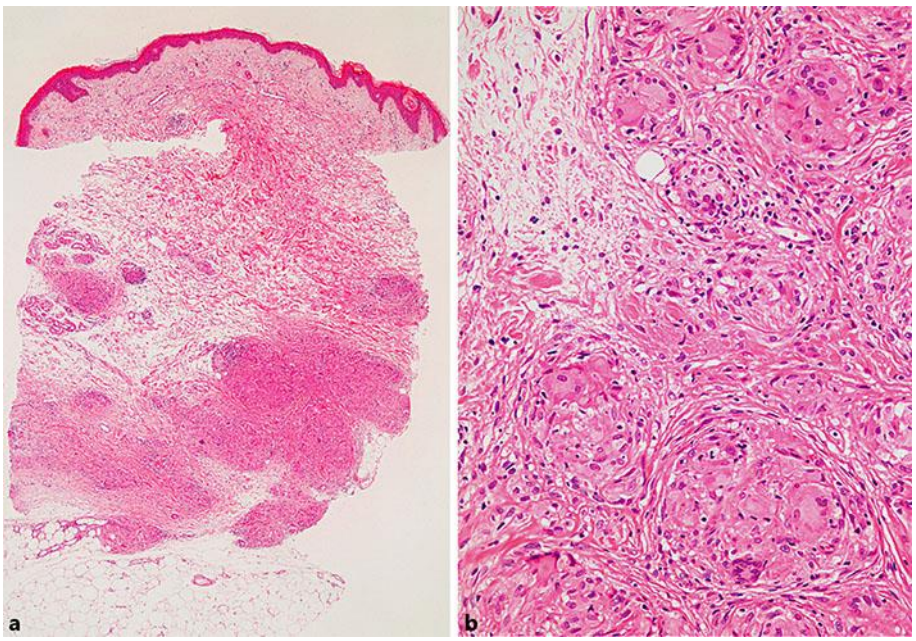

Fig. 2. **a** Histopathology showing nonnecrotic, noncaseation granuloma in the deep dermis and adipose tissue. HE. ×10. **b** A high-power view of the dermal granuloma with relatively low inflammatory cell infiltrations. HE. ×200.

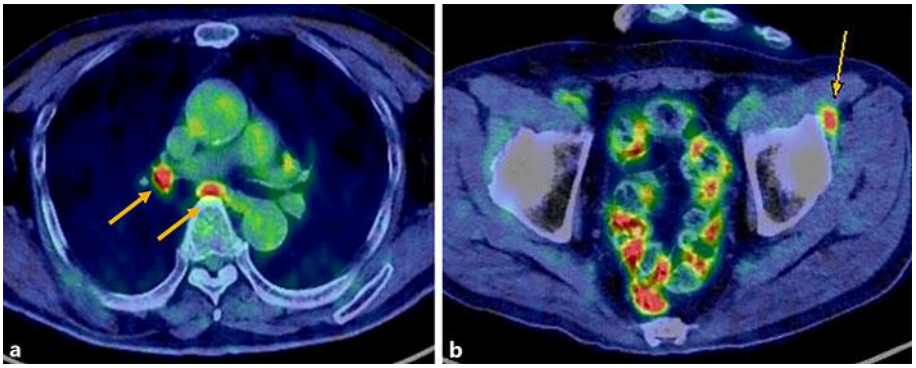

Fig. 3. Positron emission tomography screening exhibited BHL (**a**) and a solitary mass in the left gluteus minimus muscle (**b**), as indicated by orange arrows.

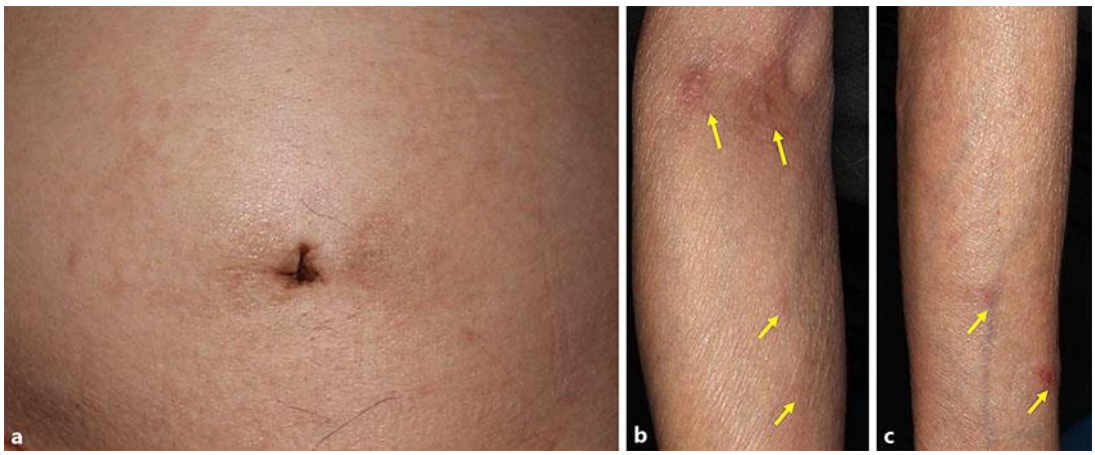

Fig. 4. Disappearance of the abdominal skin lesion with topical steroid therapy (**a**) and persisting recurrence of the forearm skin lesions along with superficial venous lines (**b**, **c**).