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Subcutaneous sarcoidosis on the digits

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Case Report

Subcutaneous sarcoidosis is a relatively rare type of specific lesion of cutaneous sarcoidosis, mainly affecting the lower extremities. We describe a case of subcutaneous sarcoidosis arising on the digits as an initial manifestation.

A 47-year-old female visited our department complaining of subcutaneous nodules on the digits without tenderness that appeared and increased in number one year previously. She had been working as a truck driver. She had type 2 diabetes and cervical cancer operated 5 years previously. Physical examination showed that multiple subcutaneous nodules were found on the first to third digits of her dominant hand (right hand) (Figure 1). Histological examination showed non-caseating epithelioid cell granulomas in the lower dermis and subcutis with multinucleated giant cells, and mononuclear cell infiltration (Figure 2,3). Detailed physical examination revealed subcutaneous nodules on the bilateral forearms, and scar sarcoidosis on the knees. Laboratory tests showed increased levels of angiotensin-converting enzyme (34.2 U/l, normal 7 to 25), and soluble Interleukin-2 receptor (883 U/ml, normal 121 to 613). Blood chemistry data showed increased levels of serum aspartate aminotransferase (44 U/L, normal 13 to 30), alanine transaminase (37 U/L, normal 7 to 23), and normal renal function. Chest X-ray showed bilateral hilar lymphadenopathy (BHL). CT scan showed enlarged bilateral hilar lymph nodes and mediastinum adenopathy. Bronchoscopic biopsy revealed non-caseating epithelioid granulomas with mononuclear cell infiltration and Zeihl-Neelsen stain was negative. Detailed examination including electrocardiogram and echocardiography excluded cardiac sarcoidosis, and ophthalmological examination did not reveal ocular sarcoidosis.

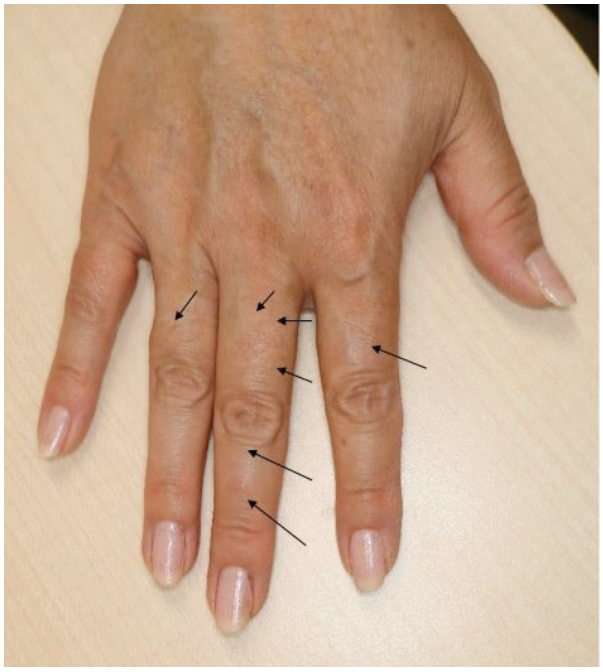

Fig. 1. Multiple subcutaneous nodules on the dorsum of the fingers.

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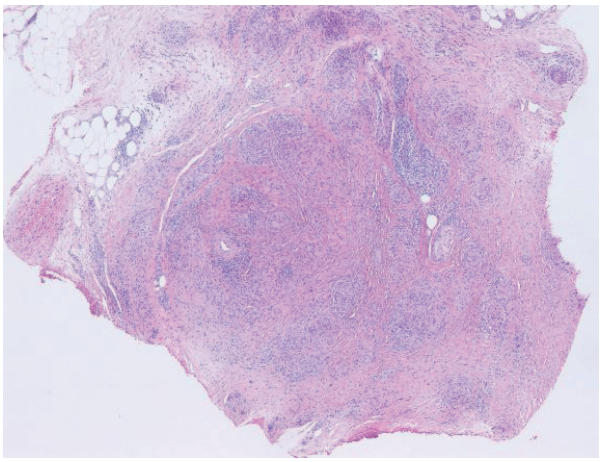

Fig. 2. A biopsy specimen from the right index finger shows non-caseating epithelioid cell granulomas with multinucleated giant cells in the lower dermis and subcutis. (H&E stain, original magnification ×200, and ×400)

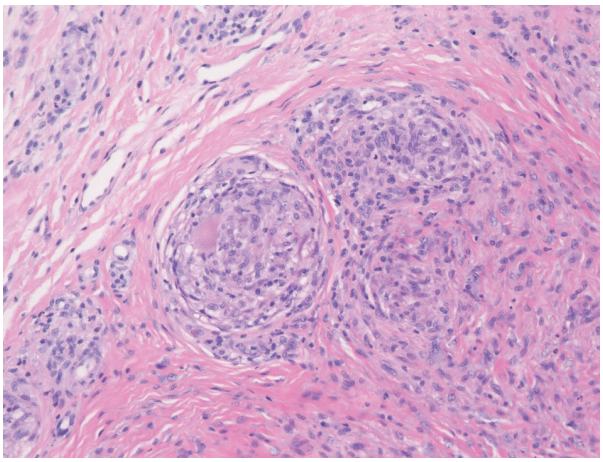

Fig. 3. A biopsy specimen from the right index finger shows non-caseating epithelioid cell granulomas with multinucleated giant cells in the lower dermis and subcutis. (H&E stain, original magnification ×200, and ×400)

The frequency of subcutaneous sarcoidosis has been estimated between 1.4 and 6% of the patients with systemic sarcoidosis (1,2). By contrast, a recent study has shown that subcutaneous sarcoidosis was observed in 11.8% (10/85) of specific cutaneous sarcoidosis cases (3). Therefore, cases of subcutaneous sarcoidosis may exist at higher frequency than was expected. According to a previous report on subcutaneous sarcoidosis, there is a female predominance and extremities are frequently involved (4). By contrast, cases of subcutaneous sarcoidosis occurring on the acral sites are rare, and only a few cases of digital occurrence have been reported to date (5-7). The previously reported cases of acral subcutaneous sarcoidosis is shown in Table 1. All cases were in around middle-aged adults, and there was no gender predominance. Dactylitis of the fingers was accompanied except for the present case. Bilateral hilar lymphadenopathy and mediastinal lymphadenopathy were observed in all cases except for 1 unknown case, suggesting that acral subcutaneous sarcoidosis is closely related to lung sarcoidosis. Other organ involvement, *i.e.* ophthalmological and cardiac sarcoidosis, were not observed, respectively.

The present case developed subcutaneous nodules on the digits as an initial manifestation, and detailed examination revealed similar subcutaneous nodules on the forearms, as well as pulmonary sarcoidosis. Our case suggests the need of histological examination when diagnosing patients presenting with acral subcutaneous nodules. Finally, our patient was a truck driver for a long time, and engaged in loading and unloading of heavy loads, and thus frequently used her hands. The present case developed subcutaneous nodules on her dominant hand, which may suggest Köbner phenomenon in sarcoidosis (8).

Table 1. Reported cases of acral subcutaneous sarcoidosis

Source	Age	Sex	Duration	Location of Lesions	Systemic Sarcoidosis
Curco N et al (5)	60	F	3 months	second finger on both hands, legs and forearms	BHL and mediastinal adenopathy
Morganroth PA et al (6)	52	M	No data	left second finger and right fourth finger, trunk and arms	BHL and mediastinal adenopathy
González-Cantero Á et al (7)	35	M	6 months	right second finger	No data
Present Case	47	F	1 year	first to third fingers, both forearms	BHL and mediastinal adenopathy

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