

Ulcerative sarcoidosis on the leg and scalp

Yasunobu Kato¹, Hideko Okabe², Michiko Hoshi², Toshiyuki Yamamoto¹

¹Department of Dermatology, Fukushima Medical University, Fukushima, Japan, ²Department of Dermatology, Hoshi General Hospital, Fukushima, Japan

Corresponding author: Prof. Toshiyuki Yamamoto, E-mail: toyamade@fmu.ac.jp

Sir,

We report herein two cases of ulcerative sarcoidosis, one of which involved a rare site, scalp.

Case 1: An 80-year-old woman was referred to the dermatology clinic, complaining of erythematous eruptions of the lower legs which had appeared nearly 30 years previously. When she underwent cataract operation, ophthalmological sarcoidosis was suspected, and she was referred to us. Physical examination showed large and small infiltrative erythematous plaques scattered on the anterior aspects of the bilateral lower legs. Biopsy revealed non-caseating granulomas with epithelioid cells in the dermis (Fig. 1a). Serum levels of angiotensin-converting enzyme (ACE) were normal (15.4 IU/L, normal: 7-25); however, serum blood urine nitrogen (27.0 mg/dl, normal: 8-22), creatinine (1.57 mg/dl, normal: 0.4-0.7), and calcium (10.4 mg/dl, normal: 8.7-10.3) levels were increased, suggesting renal sarcoidosis. Chest computed tomography (CT) scan and Gallium scintigraphy excluded bilateral hilar lymphadenopathy. During the follow-up period, she developed ischemic heart failure. Treatment with topical corticosteroid ointment was started; however, during the course, the plaque ulcerated without trauma (Fig. 1b). Thereafter, ulceration was epithelized by topical therapy without systemic prednisolone.

Case 2: A 73-year-old woman complained of shortness of breath, and examination revealed a complete atrioventricular block on electro-cardiogram and left ventricular systolic dysfunction on echocardiography. During hospitalization, she was referred to our department, complaining of skin lesions on the face, scalp and lower extremities. Physical examination revealed a large ulcer partially covered with crusts

on the scalp (Fig. 2). Furthermore, brown-reddish infiltrative plaques were scattered on the face and lower legs. Laboratory examination revealed increased levels of CRP (1.42 mg/dl) and ACE (26.1 U/L), and elevated soluble IL-2R (1290 U/ml, normal: 124-466). CT revealed bilateral hilar and mediastinum lymphadenopathy. Ophthalmological examination revealed ocular sarcoidosis, and endomyocardial biopsy revealed fibrosis without sarcoidal granulomas. A skin biopsy taken from the scalp revealed lack of epidermis, infiltration of inflammatory cells in the upper dermis, and non-caseating epithelioid cell granulomas in the dermis (Figs. 3a and 3b); however, granulomatous vasculitis was not observed. Another biopsy from the lower leg showed non-caseating epithelial cell granuloma in the dermis. The ulcers were treated with topical gentamicin sulfate ointment, which resulted in epithelization 5 months later.

Ulceration in cutaneous sarcoidosis is rarely seen, and commonly arises from pre-existing lesions. The legs are the most common sites, and a recent review collected 34 cases, 85% (29/34) of which involved the legs along with other locations including the face, arms, trunk, and genital area. Another survey of 22 Japanese patients revealed that 16 had leg ulcers, 2 had head ulcers, 1 had a buttock ulcer, and 2 had ulcers in multiple locations (unknown:1) [1]. Patients with ulcerative sarcoidosis tend to have lung and ocular lesions. Both of our patients developed ocular, lung, and cardiac sarcoidosis. Furthermore, renal sarcoidosis was suspected in Case 1. It has been reported that ulcerative sarcoidal lesions are associated with severe and active sarcoidosis. Ulcerative sarcoidosis involving the scalp is rare. Patients with scalp sarcoidosis usually have cutaneous sarcoid lesions also on sites other than the scalp [2-4].

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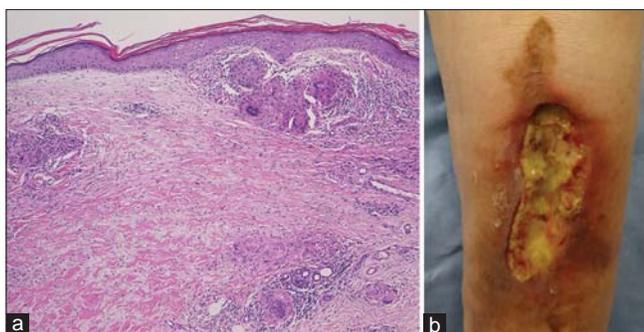


Figure 1: (a) Histological features showing epithelioid granuloma in the dermis (Hematoxylin-eosin, x100). (b) Ulceration and surrounding plaques on the shin.



Figure 2: A large ulceration with alopecia on the scalp.

In the majority of cases, scalp sarcoidosis is seen in patients with active systemic sarcoidosis. In this report, Case 2 developed cutaneous sarcoidosis on not only the scalp but also the face and lower legs. This is consistent with previously reported cases of scalp sarcoidosis.

Although previous studies suggested that granulomatous vasculitis, necrotizing granulomas, and hyaline degeneration were the possible causes of ulceration in sarcoidosis, the mechanism of ulceration in sarcoidosis remains uncertain. In our cases, fibrosis was prominent but vasculitis was not detected in the biopsied specimens, which may be due to the biopsied site. We

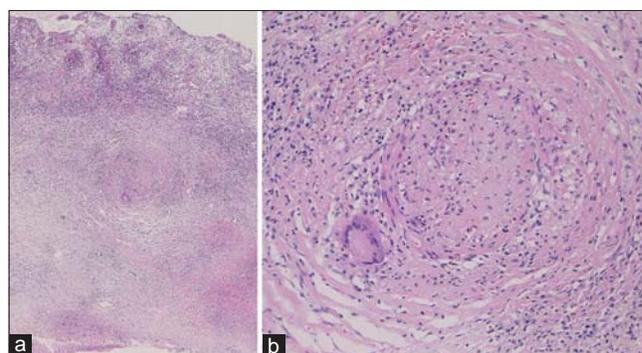


Figure 3: (a) Histological examination revealed a lack of epidermis, inflammatory cell infiltration in the upper dermis, and sarcoidal granulomas in the mid-dermis (x40). (b) Non-caseating epithelioid granulomas in the dermis (x400).

speculate that ischemic conditions may be relevant to the ulceration of sarcoidal plaques.

Consent

The examination of the patient was conducted according to the Declaration of Helsinki principles.

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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