

A Two-year-old Boy with a Depressed Area in the Groin: A Quiz

Kana OSAWA, Yukiko KINIWA* and Ryuhei OKUYAMA

Department of Dermatology, Shinshu University School of Medicine, 3-1-1 Asahi, Matsumoto, Nagano 390-8621, Japan. *E-mail: ykiniwa@shinshu-u.ac.jp

A 2-year-old boy presented with a reddish eruption with ulceration in the right axilla. The ulcer was epithelialized by topical treatment. However, a similar skin lesion subsequently developed in the left groin region and extended to the left side of the abdomen. When the patient was referred to our department, demarcated annular brownish erythema with central depression and hypopigmentation in the left

abdominal-inguinal region were observed (Fig. 1A). The central area was associated with translucent, atrophic findings. A 1-cm painful ulcer on the reddish margin was also present. The axillary skin was thin, and atrophic scarring remained. Laboratory tests were normal. Skin biopsy was performed across the annular and depressed areas (Fig. 1B).

What is your diagnosis? See next page for answer.



Fig. 1. (A) Annular brownish erythema with a hypopigmented, depressed area and ulceration. (B) Skin biopsy from the boundary of a depressed area and annular erythema. Fat necrosis and mild thickening of the septa in the depressed area, and dense perivascular and periadnexal lymphocytic infiltration in the brownish erythema. Scale bar: 500 µm (haematoxylin-eosin).

ANSWERS TO QUIZ

A Two-year-old Boy with a Depressed Area in the Groin: A Commentary

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Diagnosis: Lipodystrophia centrifugalis abdominalis infantilis

Skin biopsy revealed perivascular and periadnexal lymphocytic infiltration in the deep dermis of the annular area, and fat necrosis with mild thickening of the septa in the depressed area. Based on the clinical and pathological features, a diagnosis of lipodystrophia centrifugalis abdominalis infantilis (LCAI) was made. The ulcer was gradually epithelialized by topical corticosteroid treatment. Subsequently, another painful ulcer with erythema developed in the groin region and was treated using non-steroidal anti-inflammatory suppositories in addition to topical corticosteroids.

LCAI is a rare skin condition characterized by localized lipodystrophy that develops mainly in young children. Most cases have been reported from East Asia (1). The racial difference in the incidence rate may be due to differences in HLA alleles and skeletal structures. HLA-B51 was found in all 5 cases examined (2). Clinical features of LCAI include the development of depressed and/or atrophic areas larger than 2 cm in diameter that are surrounded by erythema and scales. In addition, 80% of cases develop in the inguinal or adjacent regions, with the remaining developing in axillary regions (1). Perivascular dermatitis and panniculitis are histopathologically observed, which are occasionally associated with lipolysis or fat necrosis. Although systemic symptoms and blood test abnormalities are rare, fever is associated in 3.6% of cases and autoantibodies are detected in 3.6% of cases (1).

The characteristics of previously reported cases are summarized in Table I. In most cases, the atrophic lesions appear during childhood and spontaneously resolve within several years. The mean age of onset was 2.5 years, 90% of cases developed before 5 years of age, and 90% ceased spontaneously before 13 years of age (1–7). Furthermore, resolution of depressed lesions was reported in approximately

60% of cases. Several treatments, including ultraviolet irradiation, topical corticosteroids, and immunosuppressants, have been sporadically successful, but no effective treatment has been established to date. Skeletal and mechanical stresses are suspected to be involved in the pathogenesis (1), but the mechanism remains to be elucidated.

Ulceration is not common in LCAI. Among 178 previous cases, 5 (2.8%), including the current case, have been reported (1–3). In the present case, the lesion extended as erythema to the lower abdomen and the inguinal region with the development of ulceration. This clinical course suggests that strong inflammation and/or fat necrosis caused localized ischaemia followed by ulceration. In addition, the localization of ulcer in the current case suggested that friction or mechanical pressure from the child's nappy is involved in the pathogenesis of lipodystrophy. Moreover, ischaemia on the borderline of 2 neighbouring angiosomes, the iliac crest and the pubic region, is a likely cause of ulcers due to damage of interlobular vessels in the subcutaneous fatty tissue (2).

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Table I. Characteristics of lipodystrophia centrifugalis abdominalis infantilis

Races	Asian: 95.5% European/European American: 4.5%
Age of onset (year)	Average: 2.5 90% percentile: 0–5
Clinical features	
Initial sites	Groin/abdomen/buttocks: 80% Axilla: 16%
Manifestations	Depression/atrophy of skin, discoloration, erythema surrounding depressed area, swelling of regional lymph nodes
Histopathological features	Dystrophic change and dense lymphocytic infiltration in adipose tissue
Clinical course	
Cessation of enlargement by 7-year-old	60%
Cessation of enlargement by 13-year-old	90%
Resolution of depressed lesions	62%
Treatments reported to be effective	Topical and/or systemic corticosteroids, pimecrolimus, tacrolimus, ultraviolet irradiation