Local Panatrophy Associated with Pain: A Rare Variant of Local Panatrophy or a New Entity?

Di-Qing Luo[1], Chang-Zheng Huang[2], Wei Shi[3], Zhuo Wang[4], Ding-Yang He[1]

[1]Department of Dermatology, The Eastern Hospital of The First Affiliated Hospital, Sun Yat-sen University, Guangzhou, Guangdong 510700, China
[2]Department of Dermatology, Union Hospital, Tongji Medical College, Huazhong University of Science and Technology, Wuhan, Hubei 430022, China
[3]Department of Dermatology, Xiangya Hospital, Central South University, Changsha, Hunan 410008, China
[4]Department of Pathology, The Eastern Hospital of The First Affiliated Hospital, Sun Yat-sen University, Guangzhou, Guangdong 510700, China

Di-Qing Luo and Chang-Zheng Huang contributed equally to this work.

To the Editor: Local panatrophy, first described by Gower in 1903 and also called panatrophy of Gowers, is an exceptionally rare disorder characterized by asymptomatic atrophy of the overlying skin as well as partial or total loss of subcutaneous tissue, and may involve the underlying muscles and bones.[1] Herein, we reported a case with such a condition associated with severe pain.

An otherwise healthy 19-year-old Chinese Han man was referred for painful atrophic patches. Two years before his presentation, an asymptomatic atrophic patch on the left posterior aspect of trunk appeared, which gradually increased in size. Five months ago, progressive lightening pain on the lesion occurred, lasting from tens of minutes to hours and attacking 3–4 times daily. Two months ago, two new red patches presented, which were located above the previous one in a vertical linear pattern, associated with severely progressive lightening pain and increased gradually in size. Warmth, movement or stimulating the lesion could trigger the attack or worsen the pain, while cooling or rest could relieve the pain. The pain had poor response to celecoxib or indomethacin alone, and mild to ibuprofen and gabapentin, aspirin combined with paroxetine, but excellent to topical compound lidocaine cream (lidocaine/prilocaine: 25 mg/25 mg/g) with 3 h of pain relief. The combination of paroxetine, aspirin, and topical compound lidocaine cream could decrease the flaring frequency initially, but lost the efficacy later. Neither his medical history nor his family history was unremarkable.

Cutaneous examination showed three atrophic patches on the posterior aspect of left trunk along spine axis. Two developing lesions (lesions 1 and 2, the subsequent), distributed on the chest, were mild-atrophic erythema (6.5 cm × 5.0 cm and 5.5 cm × 5.5 cm, respectively), with irregularly ill-defined bounder and telangiectasis [Figure 1a]. The advanced one (lesion 3, the initial), localized on the waist, was an irregularly sunken pigmented patch (about 9.0 cm × 6.5 cm), with wasted skin and underlying tissues [Figure 1a]. All three lesions, especially lesion 3, had marked tenderness and hyperesthesia, with visible subcutaneous veins. However, no sclerosis or binding down the deeper tissue presented. The skin over the lesions was freely mobile over the subjacent tissues. Neither increased temperature and blood pressure nor flaring erythema presented during the pain attacking. No other parts involved. By B-ultrasonic scan, the thicknesses of the epidermis plus dermis/subcutaneous tissue for lesions 1, 2, and 3 were 2.5 mm/4.2 mm, 3.0 mm/4.2 mm, and 2.4 mm/2.4 mm, respectively, while those of normal skin on the mirror parts of opposite side were 4.2 mm/4.6 mm, 4.3 mm/4.6 mm, and 3.9 mm/3.9 mm, respectively. Magnetic resonance imaging revealed atrophic both dermis and subcutaneous tissue over the lesions, with preference in lesion 3, and the underlying muscles, skeletons, and spine nerve showed normal. Biopsies from the center of both lesions 1 and 3 showed pigmented basal layer of the epidermis; decreased epidermis, dermis, and subcutaneous tissue including fat tissue; and slight perivascular lymphocytic infiltrate in dermis; with predominance in lesion 3 [Figure 1b and 1c]. No inflammatory infiltrates were presented in the skin nerve [Figure 1b]. The biopsy from the perilesion showed mild inflammatory infiltrate [Figure 1d]. Aldehyde-fuchsin stain revealed that specimens from lesion 1 and from perilesion 3 showed fragmented and rarefied elastic fibers in the dermis, but only mild alterations in lesion 3 except for evident decrease of dermis and subcutaneous tissue. Autoantibodies and antinuclear antibody and serology for syphilis and lyme borreliosis were negative. During 5-year follow-up, lesion 3 progressed slowly in size while the rests changed mildly, the pain had moderate self-alleviation, and no new lesions occurred.

Based on the clinical and pathological alterations, we considered that the present case was a variant of local panatrophy associated with pain rather than a new entity. Of course, we could not absolutely exclude the possibility that the present condition

Address for correspondence: Dr. Di-Qing Luo, Department of Dermatology, The Eastern Hospital of The First Affiliated Hospital, Sun Yat-sen University, 183 Huangpu Rd. E., Guangzhou, Guangdong 510700, China
E-Mail: luodq@mail.sysu.edu.cn

Access this article online

Quick Response Code:

Website: www.cmj.org

DOI: 10.4103/0366-6999.216400

Received: 20-07-2017 **Edited by:** Xin Chen
How to cite this article: Luo DQ, Huang CZ, Shi W, Wang Z, He DY. Local Panatrophy Associated with Pain: A Rare Variant of Local Panatrophy or a New Entity? Chin Med J 2017;130:2515-6.

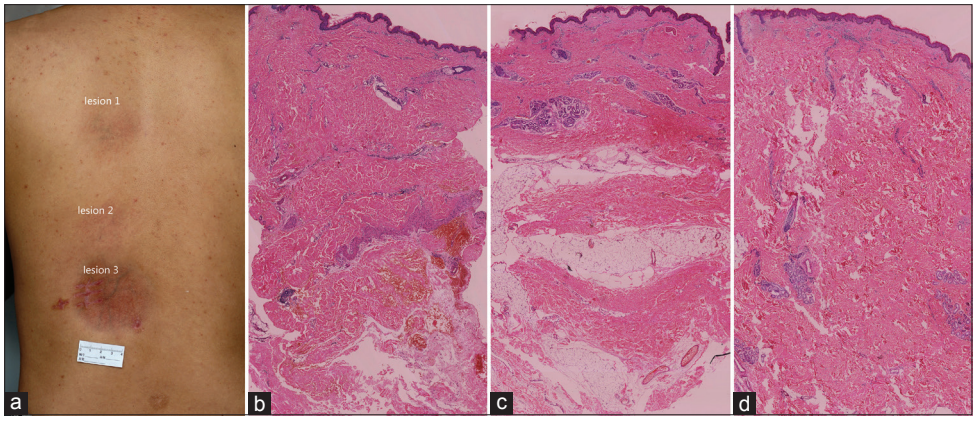

Figure 1: Representative images of the patient. (a) Three atrophic patches distribute on the posterior aspect of left trunk along spine axis, with irregularly ill-defined bounder, telangiectasis, and visible subcutaneous veins (lesions 1 and 2: developing; lesion 3: advanced). Biopsies from the center of both lesions 1 (b) and 3 (c) showed decreased underlying epidermis and subcutaneous tissue, and slight perivascular lymphocytic infiltrate, with predilection of lesion 3; while that from the perilesional area showed mild inflammatory infiltrate (d) (H & E, original magnification, ×25).

represented a new clinicopathologic entity. The possible reasons for the different presentations of elastic fibers in advanced lesion, developing lesion, and peri-lesions were that most elastic fibers in the dermis of advanced lesion disappeared or lost while elastic fibers in developing lesion and perilesional area were just damaged alone, implying that the elastic fibers were damaged in early stage and most damaged fibers disappeared later. However, the reasons why the pain had poor response to therapeutics remain unknown.

The differential diagnoses mainly include anetoderma, mid-dermal elastolysis, focal dermal hypoplasia, lupus panniculitis, and localized scleroderma. Based on their clinical and histological features,[2-5] making a correct diagnosis is not difficult.

Declaration of patient consent

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.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Sakamoto T, Oku T, Takigawa M. Gowers' local panatrophy. Eur J Dermatol 1998;8:116-7.
2. Chisholm C, Miedler J, Etufugh CN, Ghali F, Cockerell CJ. Unusual and recently described cutaneous atrophic disorders. Int J Dermatol 2011;50:1506-17. doi: 10.1111/j.1365-4632.2011.04938.x.
3. Lewis KG, Bercovitch L, Dill SW, Robinson-Bostom L. Acquired disorders of elastic tissue: Part II. Decreased elastic tissue. J Am Acad Dermatol 2004;51:165-85. doi: 10.1016/j.jaad.2004.03.016.
4. Gambichler T. Mid-dermal elastolysis revisited. Arch Dermatol Res 2010;302:85-93. doi: 10.1007/s00403-009-1004-0.
5. Valanciene G, Jasaitiene D, Valiukeviciene S. Pathogenesis and treatment modalities of localized scleroderma. Medicina (Kaunas) 2010;46:649-56.