Case Reports in Dermatology

Case Rep Dermatol 2012;4:66–71
DOI: 10.1159/000337636

Published online:
March 23, 2012

ISSN 1662–6567
www.karger.com/cde

Cutaneous Small Vessel Vasculitis Accompanied by Pustulosis Palmaris et Plantaris

Motoko Kosaka Tokue Kato Seiji Kawana

Department of Dermatology, Nippon Medical School, Tokyo, Japan

Key Words
Cutaneous small vessel vasculitis · Pustulosis palmaris et plantaris · Pustulotic arthro-osteitis · Apical periodontitis · Radicular cyst · Focal infection

Abstract
We present the case of a 64-year-old woman who has suffered from pustulosis palmaris et plantaris for 10 years. At the first examination, many erythematous lesions with purpura, blood crusts, and blisters were present in the lower legs and dorsum of the feet. Painful swelling in the sternal region and dorsal pain were also noted. Elevation of the CRP and myogenic enzyme levels, and liver and renal dysfunctions were noted on blood testing. Histopathologically, leukocytoclastic vasculitis was noted in small blood vessels in the whole dermal layers, and deposition of IgM and C3 in the vascular wall was detected by the direct immunofluorescence techniques. Based on these findings, cutaneous small vessel vasculitis was diagnosed. Because the patient complained of a toothache during the clinical course, an X-ray examination was performed. On pantomography, a radicular cyst and apical periodontitis were noted. The tooth symptoms changed with exacerbation and remission of the skin symptoms. These findings indicate that odontogenic infection is very likely to be a cause of cutaneous small vessel vasculitis in a manner similar to pustulosis palmaris et plantaris.

Introduction

Immune complex-induced type 3 allergy is thought to be involved in the development of cutaneous small vessel vasculitis, and focal infection is assumed to be a cause. The association of pustulosis palmaris et plantaris with odontogenic infection has also been investigated. We present a case of cutaneous small vessel vasculitis accompanied by pustulosis palmaris et plantaris and suspected involvement of odontogenic infection.

Motoko Kosaka

Department of Dermatology, Nippon Medical School
1-1-5 Sendagi, Bunkyo-ku, Tokyo 113-8603 (Japan)
Tel. +81 3 3822 2131, E-Mail s7036@nms.ac.jp

Case Reports in Dermatology
Case Rep Dermatol 2012;4:66–71
DOI: 10.1159/000337636
Published online: March 23, 2012

ISSN 1662–6567
www.karger.com/cde

Case Report

We present the case of a 64-year-old woman who has suffered from pustulosis palmaris et plantaris for 10 years. She sometimes noticed a toothache, but she left this untreated. At the first examination, many erythematous lesions with purpura, blood crusts, and blisters were present in the lower legs and dorsum of the feet (fig. 1a, b). Palpable purpura with a diameter of 2–5 mm were diffusely present in the lower extremities. Pustules and thick yellow scales were present on the soles (fig. 1c), and erythemas were present on the palms. Painful swelling in the sternal region and dorsal pain were also noted.

Blood tests indicated elevated GOT, 165 IU/l; GPT, 85 IU/l; LDH, 414 IU/l; γ-GTP, 160 IU/l; CPK, 6,322 IU/l; T-Bil, 2.1 mg/dl; BUN, 34.2 mg/dl; Cre, 1.35 mg/dl; CRP, 34.54 mg/dl; IgA, 603 mg/dl; IgE, 1,315 mg/dl; and ASO, 189.0 IU/ml. The level of blood coagulation factor XIII was slightly reduced to 66% (normal range >70%). WBC, ANA, RF, PR3-ANCA, MPO-ANCA, cryoglobulin, PT, and APTT were within the normal ranges. In urinalysis, protein was 2+ and blood was 3+.

Histopathologically, a purpuric erythema in the lower leg showed dense infiltration of neutrophils and lymphocytes in the upper dermis, and leukocytoclastic vasculitis characterized by infiltration of neutrophils, nuclear dust, fibrinoid deposits, and erythrocyte extravasation in small blood vessels of the upper and lower dermis (fig. 1d, e).

Direct immunofluorescence indicated granular deposition of IgM and C3 in the walls of small blood vessels. Epidermal thickening and spongiform pustules were present on the toe, showing histological features of pustulosis palmaris et plantaris.

Chest CT indicated a large volume of bilateral pleural effusion (fig. 2a). Bacterial and fungal culture tests for the pleural effusion were negative. Bone scintigraphy showed marked accumulation in the sternum, sternoclavicular joints, sternocostal joints, lower costal ends, and costal cartilages.

There were no findings that indicated angiitis or focal infections in other organs in a thorough examination, including MRI, CT, Ga scintigraphy, echography, and otolaryngological and ophthalmological examinations. Because the patient complained of a toothache during the clinical course, an X-ray examination was performed. On pantomography, a radicular cyst was noted in the right lower canine, and apical periodontitis was present in the right lower first molar. Moreover, alveolar bone was resorbed in one half to one third of the root length in the right lower central incisor, canine, second molar, and left lower lateral incisor to the first premolar, leading to a diagnosis of moderate to severe chronic periodontitis (fig. 2b).

The skin eruptions and swelling of the lower limbs were relieved after 1 week with rest, treatment with a non-steroidal anti-inflammatory drug, and topical steroid application. Liver and renal functions, CPK level, and urinalysis findings normalized. However, formation of purpura and retention of pleural effusion continued, and the CRP level remained elevated. Therefore, minocycline hydrochloride was administered at a dose of 200 mg/day, and pleural effusion decreased after 1 week. Toothache and purpura lesions were improved after 4 weeks, but some pustules in the palmoplantar area and weak sternal pain still remained.

Discussion

This case showed concomitant development of pustulosis palmaris et plantaris, pustulotic arthro-osteitis, and cutaneous small vessel vasculitis. The retention of pleural effusion may have been due to pleuritis or spread of adjacent osteoarthritis to the pleura that developed concomitantly with pustulosis palmaris et plantaris. Fernandez-Campillo and Garcia-Pachon [1] reported a case showing similar symptoms.

Immune complex-induced type 3 allergy is thought to be involved in the development of cutaneous small vessel vasculitis, and focal infection is assumed to be a cause [2–4]. Odontogenic infection may be one of the focal infections. Misago et al. [5]

Case Rep Dermatol 2012;4:66–71
DOI: 10.1159/000337636
Published online: March 23, 2012

ISSN 1662–6567
www.karger.com/cde

described a case of small vessel vasculitis that remitted with treatment of periodontitis, and Tahmassebi et al. [6] reported a case in which Henoch-Schönlein purpura occurred during treatment of apical periodontitis. The association of pustulosis palmaris et plantaris with odontogenic infection has also been investigated. Of 469 patients with pustulosis palmaris et plantaris, those with focal infections accounted for 37%, including odontogenic focal infection in 12% [7]. In the dental field [8], apical periodontitis was found in 50 of 52 cases of pustulosis palmaris et plantaris (96%), and dental treatment was effective in 70% of these cases.

In the present case, apparent apical periodontitis and a radicular cyst were found on pantomography. The tooth symptoms changed with exacerbation and remission of the skin symptoms. According to these findings, we suspect that odontogenic infection was involved in the development of cutaneous small vessel vasculitis and the exacerbation of pustulosis palmaris et plantaris and pustulotic arthro-osteitis. Exclusion of the odontogenic infection and longer follow-up of this patient would be needed to confirm the influence of odontogenic infection on vasculitis and pustulosis palmaris et plantaris.

Case Reports in Dermatology
Case Rep Dermatol 2012;4:66–71
DOI: 10.1159/000337636
Published online: March 23, 2012

ISSN 1662–6567
www.karger.com/cde

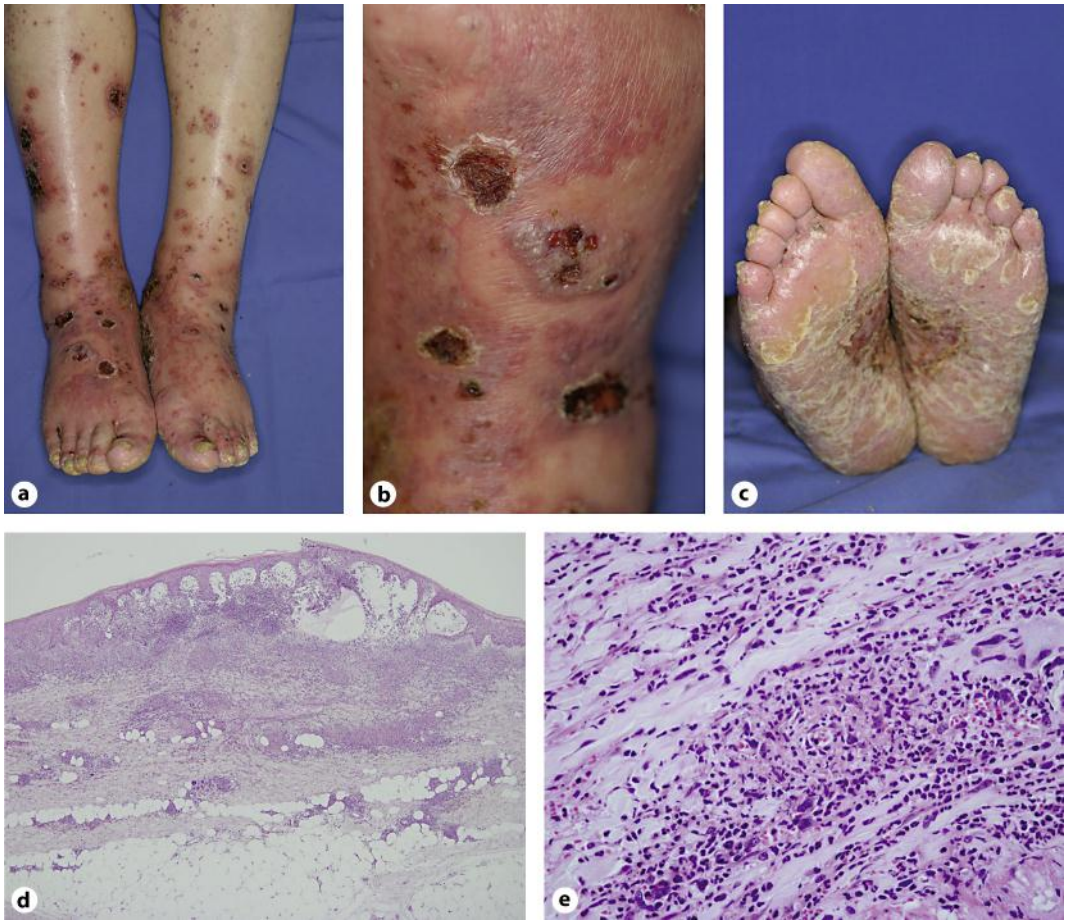

Fig. 1. Skin findings on the first examination and histopathological findings. **a**, **b** There were many erythematous lesions of 6–10 mm in diameter with crusted ulcer, pustule, blister, and blood blister. Palpable purpura of 2–5 mm in diameter was diffusely present. **c** Pustules and thick yellow scales were present on the soles. **d** Blisters were present under the epidermis. Dense infiltration of inflammatory cells containing numerous neutrophils and lymphocytes was noted in the upper dermis and the walls of small blood vessels in the upper to lower dermal layers (HE staining, ×40). **e** Skin biopsy specimen showed leukocytoclastic vasculitis characterized by infiltration of neutrophils, nuclear dust, fibrinoid deposits, and erythrocyte extravasation in and around small blood vessels (HE staining, ×400).

Case Rep Dermatol 2012;4:66–71
DOI: 10.1159/000337636
Published online: March 23, 2012

ISSN 1662–6567
www.karger.com/cde

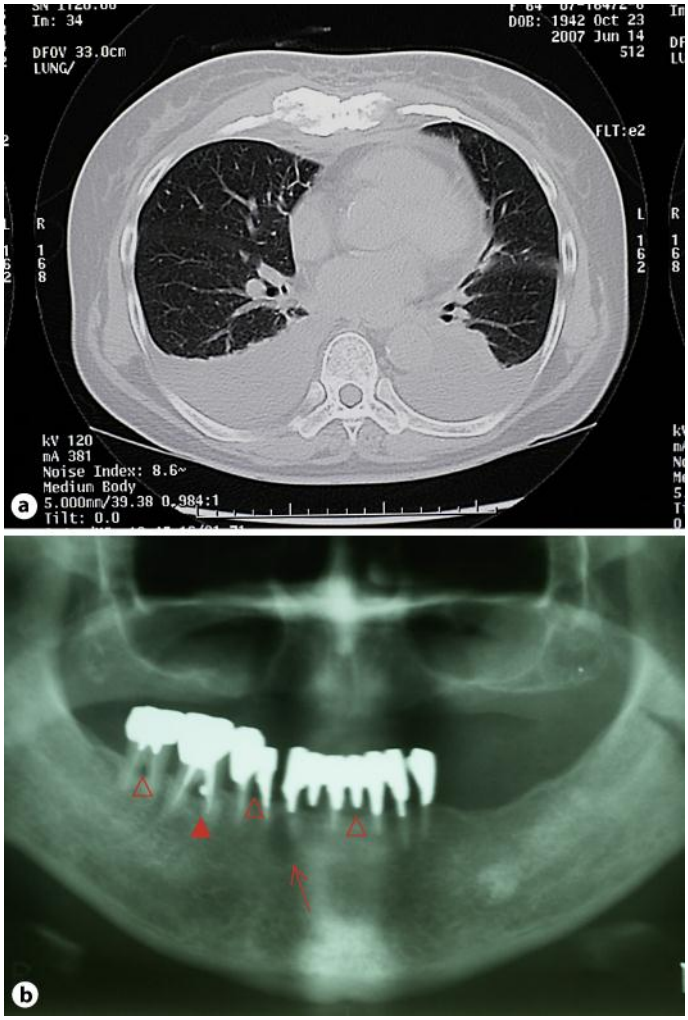

Fig. 2. **a** Chest CT showed a large volume of bilateral pleural effusion. **b** Pantomography showed a radicular cyst (→) in the right lower canine and apical periodontitis (▲) in the right lower first molar, with alveolar bone resorption (△) noted in the right lower central incisor, canine, second molar, and left lower lateral incisor to the first premolar.

References

1 Fernandez-Campillo J, Garcia-Pachon E: Pleural effusion associated with the SAPHO syndrome. Chest 2001;120:1752.

2 Russell JP, Gibson LE: Primary cutaneous small vessel vasculitis: approach to diagnosis and treatment. Int J Dermatol 2006;45:3–13.

3 Carlson JA, Chen KR: Cutaneous vasculitis update: small vessel neutrophilic vasculitis syndromes. Am J Dermatopathol 2006;28:486–506.

4 Lotti T, Ghersetich I, Comacchi C, Jorizzo JL: Cutaneous small-vessel vasculitis. J Am Acad Dermatol 1998;39:667–687.

5 Misago N, Yonekura N, Kuroiwa T, Yamanaka K, Narisawa Y: Simultaneous occurrence of reactive angioendotheliomatosis and leukocytoclastic vasculitis in a patient with periodontitis. Eur J Dermatol 2008;18:193–194.

Case Rep Dermatol 2012;4:66–71
DOI: 10.1159/000337636

Published online:
March 23, 2012

ISSN 1662–6567
www.karger.com/cde

6 Tahmassebi JF, Paterson SA: Development of acute Henoch-Schönlein purpura subsequent to endodontic treatment. Int J Paediatr Dent 2007;17:217–222.

7 Akiyama T, Seishima M, Watanabe H, Nakatani A, Mori S, Kitajima Y: The relationships of onset and exacerbation of pustulosis palmaris et plantaris to smoking and focal infections. J Dermatol 1995;22:930–934.

8 Ueda Y, Shiota S: Clinical studies on dental focal infection and interrelationship with skin disease of pustulosis palmaris et plantaris. Jpn J Oral Diag/Oral Med 1991;4:464–468.