

Case Presentation

A case of diffuse Gougerot and Blum purpuric pigmented dermatosis

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Abstract

Purpuric and pigmented lichenoid dermatitis of Gougerot and Blum is a form of purpuric pigmented dermatosis. These entities are rarely described but are likely underdiagnosed. Herein, we present a patient with this condition that is unusual in its diffuse nature and its delayed diagnosis after twenty years of evolution.

Introduction

Purpuric pigmented dermatosis (PPD) is a group of benign dermatoses whose common feature is the dermal extravasation of erythrocytes and macrophages loaded with hemosiderin.¹ Their pathophysiology is poorly understood. This group of conditions mainly affects adults, but can also occur in children.^{2,3} No association with systemic illness has been reported.¹ The diagnosis is mostly clinical and histology can be of aid in more difficult cases.^{1,4} Efficacious treatment has not clearly been determined; clearing may occur but relapses are frequent.

PPD is divided into five subcategories, which form a clinical spectrum. The most common one is progressive pigmented purpura, Schamberg disease; its main clinical features are "cayenne pepper" petechiae, which form brick-red patches on the lower limbs.¹ Majocchi purpura annularis telangiectasia is characterized by annular macular plaques of the legs. The initial peripheral telangiectasias become pigmented over time and surround an atrophic center.¹ Eczematoid-like purpura of Doucas and Kapetanakis begins on the legs and rapidly spreads to the trunk, forming very itchy purpuric and slightly scaly patches.¹ Lichen aureus is described as a solitary gold or brown, usually in a single patch.¹ Pigmented purpuric lichenoid dermatosis of Gougerot and Blum (PPLD) is a rarely reported form of PPD that mostly involves the lower limbs. Its visible and pruritic nature can be distress-

ing to patients.¹ We report a patient with diffuse PPD of Gougerot and Blum, which had been present for twenty years prior to diagnosis.

Case Synopsis

A 46-year-old woman was referred to our dermatologic clinic for a dermatosis that had been developing for about twenty years. Initially the lesions were present on the legs and wrists, but after about fifteen years of evolution, they spread to the thighs, abdomen, and upper limbs. Mild pruritus had led to prior treatment with potent topical corticosteroids, which failed to improve her condition.

Clinical examination revealed reticulated, purpuric papules with mild hyperkeratosis ([Figure 1-3](#)) of the abdomen, thighs, and upper limbs. The older lesions on the legs were pigmented. Head and neck were spared and there was no clinical involvement of mucosal membranes.

Skin biopsy showed interface dermatitis with vacuolar changes and apoptosis of basal membrane keratinocytes. There was a focal superficial perivascular infiltration of the dermis, associated with erythrocyte extravasation and hemosiderin deposition. The latter was accentuated after Perls staining ([Figures 4 A-B](#)).

These clinical and histological findings led to the diagnosis of Gougerot and Blum purpuric and pigmented lichenoid dermatitis, a rare form of PPD. Incidentally, a monoclonal lambda IgA peak of 5.6g/L was present, which was deemed of undetermined significance and unrelated after hematology consult evaluation. There was no argument for multiple myeloma during the subsequent hematological follow-up.

Subcutaneous methotrexate at a dose of 15mg per week was prescribed as symptomatic treatment for the patient's pruritus. It was discontinued after six months owing to primary ineffectiveness; subsequent treatments

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Figure 1. Reticulated papular purpuric lesions with mild hyperkeratosis of the left arm.



Figure 3. Purpuric ovoid lesions on the medial aspect of the right wrist.



Figure 2. Reticulated purpuric erythematous lesions on both thighs associated with pigmented ovoid lesions of both legs.

with rutoside, ascorbic acid, and colchicine were equally unsuccessful.

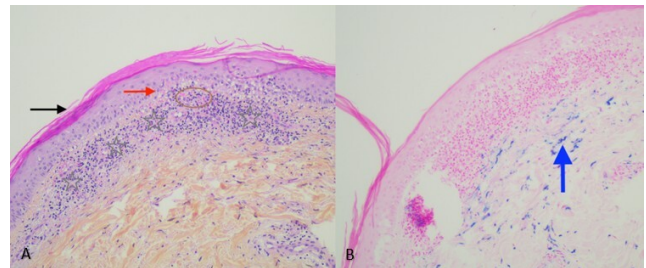


Figure 4. A) HPS x40 slide showing parakeratotic stratum corneum, mildly spongiotic epidermis, interface dermatitis, and focal superficial perivascular infiltrate associated with erythrocyte extravasation. B) Perls stain showing dermal hemosiderin deposition in blue.

Discussion

Purpuric and pigmented lichenoid dermatitis of Gougerot and Blum is a rare form of PPD and affects mostly elderly men.¹ Reddish-brown lichenoid papules develop on the legs and tend to coalesce into plaques.⁵ Evolution is chronic and can be associated with mild pruritus.¹ Dermoscopy shows oval red spots associated with erythrocytes and an orange to brown background.⁴

Diagnosis on clinical features alone can be challenging, as it may resemble Kaposi sarcoma, vasculitis, traumatic purpura, or mycosis fungoides.^{1,4} Histologically, red blood cells are extravasated into the dermis, forming the deposition of hemosiderin characteristic of PPD. Thus, Perls stain is of crucial help to reach a diagnosis. Other histological findings include a perivascular lichenoid infiltrate composed of T lymphocytes.^{3,4}

In our case, the rash on the legs and the presence of well-defined erythematous papules with lichenoid histology was consistent with a diagnosis of PPLD. However, plaques of the trunk and upper limbs are more uncharacteristic.^{1,5} Such a widespread clinical manifestation has only been described in a handful of case reports before, mostly in women.^{3,6} Eczematoid-like purpura of Doucas and Kapetanakis, which frequently expands from the lower limbs to the trunk and upper limbs, was deemed less likely in the absence of blurred plaques, only mild pruritus, and minimal spongiotic histology.

Skin biopsy was the key factor, which allowed diagnosis of PPLD in this unusual clinical presentation. This atypical form may have led to the considerable delay in diagnosis in our patient, whose dermatitis had been present for over two decades prior to her visit in our center.

Causes of PPD are still unknown. Venous insufficiency, local infections, and certain drugs, such as oral antidiabetics and benzodiazepines, may play a role in PPD development.⁷⁻⁹

Although most cases of PPD are idiopathic, some have been associated with systemic pathologies such as autoimmune diseases or hematological neoplasia.¹ Purpuric and pigmented lichenoid dermatitis of Gougerot and Blum, like PPD in general, poses a therapeutic challenge. In the absence of standardized treatment, many different approaches have been tried.¹⁰

Topical corticosteroids can help reduce pruritus,¹ but are generally regarded as lacking in efficacy.^{3,6,10} Phototherapy (narrowband UVB and photodynamic therapy) and pentoxifylline, which is no longer available in France,

seem to be interesting therapeutic approaches. They have proved relatively high efficacy in some cases^{1,6,10} and come with fewer side effects^{6,7,10} than immunomodulating and immunosuppressive agents (colchicine, dapsone, methotrexate).^{7,10} Calcineurin inhibitors have been helpful in eczematoid and lichenoid forms.¹⁰

Other cases have been reportedly treated with vitamin C or rutoside/rutin with a focus on antioxidant effects or decreasing capillary fragility with variable results.^{1,10}

Kimak et al thoroughly reviews mechanisms and treatment options, but the absence of standardized protocols leads to a variability in results.¹⁰ Finally, a watchful waiting approach is also a valid therapeutic option in this benign pathology, provided patients are asymptomatic.¹⁰

Conclusion

Purpuric pigmented dermatosis is a rare, benign condition in which histology can be of help in cases of uncharacteristic clinical presentations. Our patient with an extensive purpuric and pigmented lichenoid dermatitis of Gougerot and Blum capillaritis exhibits the occasional difficulty in diagnosis of forms of PPD. Treatment may be optional in asymptomatic cases but poses a therapeutic challenge in symptomatic patients.

Potential conflicts of interest

The authors declare no conflict of interest.

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