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Journal

Dermatology Online Journal, 31(3)

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Publication Date

2025

DOI

10.5070/D331365372

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Peer reviewed

Asymptomatic telangiectatic macules on the trunk and proximal extremities

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Keywords: collagen, microangiopathy, telangiectasias, vasculopathy

Dermatology Online Journal

Vol. 31, Issue 3, 2025

Abstract

Cutaneous collagenous vasculopathy is a rare microangiopathy that presents with asymptomatic telangiectasias and distinct histopathological features, including hyaline material deposition in vessel walls. Diagnosis requires biopsy and differentiation from other telangiectatic disorders. Current treatment options are limited, but increased awareness may improve recognition and management of this rare condition. Herein, we describe an unusual presentation of cutaneous collagenous vasculopathy as asymptomatic, pink, mat-like telangiectatic macules on the trunk and proximal extremities of a 62-year-old woman. Histopathologic evaluation revealed dilated superficial dermal vessels with hyaline thickening of the vessel walls, consistent with a diagnosis of cutaneous collagenous vasculopathy.



Figure 1. Clinical presentation of cutaneous collagenous vasculopathy. **A)** Mat-like telangiectasias on the trunk of a 62-year-old woman (black arrowheads). **B)** A 10mm discrete, round, telangiectatic macule on the trunk.

telangiectatic conditions, emphasizing the importance of histopathologic evaluation for accurate diagnosis.

Introduction

Cutaneous collagenous vasculopathy (CCV) is a rare microangiopathy characterized by diffuse, asymptomatic telangiectasias that typically begin on the lower extremities.¹ First described in 2000,² CCV predominantly occurs in middle-aged and elderly individuals, with a slight female predominance.¹ Clinically, it resembles other telangiectatic disorders, such as generalized essential telangiectasia, but is distinguished by its unique histopathologic features, including dilated superficial dermal blood vessels with thickened walls related to hyaline material deposition and reduplication of the basement membrane.² Although CCV is considered rare, it may be underdiagnosed because of its similarity to other

Case Synopsis

We report a 62-year-old woman who presented with a one-year history of asymptomatic pink lesions on her trunk and proximal extremities. The lesions first presented on the right breast, then spread to involve the left breast, back, chest, arms, and legs. Her past medical history was significant for hypertension, hyperlipidemia, and diabetes mellitus. Her current medications included valsartan and simvastatin. Her only new medication was brinzolamide eyedrops. Physical examination showed numerous 2mm to 10mm discrete, round telangiectatic macules distributed over the trunk and proximal extremities (**Figures 1A-B**), which partially blanched. There was no conjunctival or mucosal involvement.

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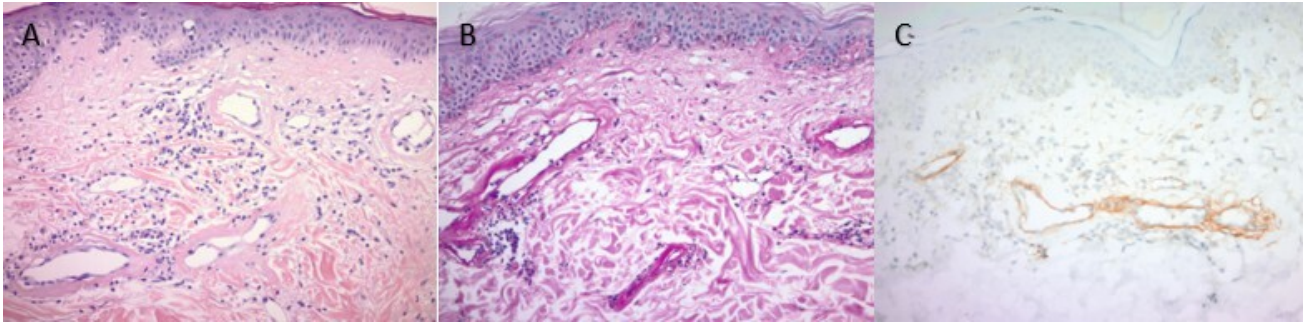


Figure 2. Punch biopsy specimen. **A)** Dilated superficial vessels with pink hyalinized thickened vessel walls (hematoxylin eosin stain, original magnification 200X). **B)** Periodic acid-Schiff-positive material around vessel walls (Periodic acid-Schiff, original magnification, 200X). **C)** Immunohistochemistry with anti-collagen 4-antibody revealing collagen accumulation around vessel walls (collagen 4 marker, original magnification, 200X).

Histopathologic examination of a 4mm punch biopsy from the back demonstrated dilated vessels in the superficial dermis with vessel wall thickening owing to the deposition of eosinophilic hyaline material and reduplication of the basement membrane (**Figure 2A**). The hyaline material stained positive for periodic acid-Schiff (**Figure 2B**) and collagen 4 (**Figure 2C**). These clinicopathologic findings are consistent with CCV. Our patient was treated once with pulsed dye laser therapy, but the lesions persisted at follow-up. She declined further treatment.

Case Discussion

Cutaneous collagenous vasculopathy (CCV) is a rare, benign primary cutaneous microangiopathy that clinically resembles generalized essential telangiectasia, but with distinct histopathology and unique ultrastructural features. Clinically, patients present with diffuse asymptomatic telangiectasias, which often begin on the lower extremities and ascend to involve the trunk and upper extremities, while sparing the nails and mucosa.¹ However, some cases of CCV have been described as first presenting on the breasts or trunk (**Figures 1A-B**) then spreading progressively to the extremities, as seen in our patient.³ The present case was also somewhat unusual in that all the lesions occurred as round, mat-like telangiectasias instead of “spider-like” telangiectasias typically seen in CCV and other telangiectatic disorders. Cases of CCV have been primarily described in middle-aged and elderly individuals and may exhibit a slight predominance in females.¹ Many patients take at least one medication regularly or have concurrent hypertension, cardiovascular disease, diabetes mellitus, or autoimmune disease.⁴ There have been several reports of CCV cases associated with autoimmune stigmata, including positive serologic anti-endothelial cell antibody assay, lupus anticoagulant, and anti-ribonucleoprotein antibodies.⁵ However, the relationship between CCV and connective tissue disease has not yet been elucidated and laboratory workup for connective tissue disease markers in CCV patients is not generally recommended.⁵

The pathogenesis of CCV is unknown, but one proposed mechanism is that repeated endothelial cell injury may activate veil cells, leading to defective collagen deposition in cutaneous blood vessel walls.⁴ CCV can be challenging to differentiate clinically from other telangiectatic disorders and biopsy is necessary for diagnosis. Histopathologically, CCV is characterized by its unique histological appearance of mildly dilated blood vessels in the superficial dermis with hyaline thickening of the vessel walls owing to collagen deposition and reduplication of the basement membrane (**Figure 2A**).² Sparse inflammation may be seen in the surrounding dermis. The hyaline material stains positive for periodic acid-Schiff (**Figure 2B**), collagen 4 (**Figure 2C**), fibronectin, and laminin; it is diastase resistant and negative for elastin and smooth muscle actin.² Endothelial cells of telangiectatic vessels express *CD31* and *CD34*, as well as von Willebrand factor.² On electron microscopy, veil cells surrounded by abnormal collagen (Luse bodies) in outer vessel walls may also be seen.² Current treatment options for CCV are limited, but pulsed dye laser therapy has led to successful treatment of telangiectatic lesions in some patients.¹

Conclusion

CCV is a rare entity with fewer than 50 cases reported in the literature to date and is likely underrecognized. Proper diagnosis requires histopathologic evaluation and clinical correlation to differentiate it from other telangiectatic disorders like generalized essential telangiectasia and increased awareness may lead to better recognition, understanding of its pathogenesis, and treatment.

Potential conflicts of interest

The authors declare no conflicts of interest.

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