Volume 22 Number 4
April 2016

Case presentation

Primary cutaneous angioplasmocellular hyperplasia

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Dermatology Online Journal 22 (4): 6

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Abstract

Angioplasmocellular hyperplasia is a rare clinical condition with blood vessel proliferation and a reactive plasma cell infiltrate. To the best of our knowledge fewer than 20 cases of cutaneous angioplasmocellular hyperplasia have been published in English literature. We report a case of a 65-year-old man who presented with a long standing asymptomatic flesh colored ulcerated nodule on the back. Histopathological examination revealed a dermal vascular proliferation with polyclonal plasma cell infiltration. A diagnosis of angioplasmocellular hyperplasia was established. This entity is rare and we would like to emphasize the importance of clinico-pathological correlation to differentiate it from various other conditions of cutaneous plasma cell infiltration.

Introduction

Angioplasmocellular hyperplasia is a rare clinical condition with blood vessel proliferation and a reactive plasma cell infiltrate. It was first reported in two adults as solitary nodules on the trunk [1]. Primary cutaneous angioplasmocellular hyperplasia has been proposed as a distinct entity composed of plasma cell infiltrates and cutaneous vascular proliferation [2].

Case synopsis

A 65-year-old farmer presented with a slowly growing lesion on his back for a duration of 15 years that had suddenly increased in size over 6 months prior to presentation. During these 6 months he reported multiple episodes of spontaneous mild bleeding from the nodule. There was no history of trauma or insect bite prior to the onset. There was no history of any hepatic dysfunction, diabetes mellitus, hypertension, or tuberculosis.

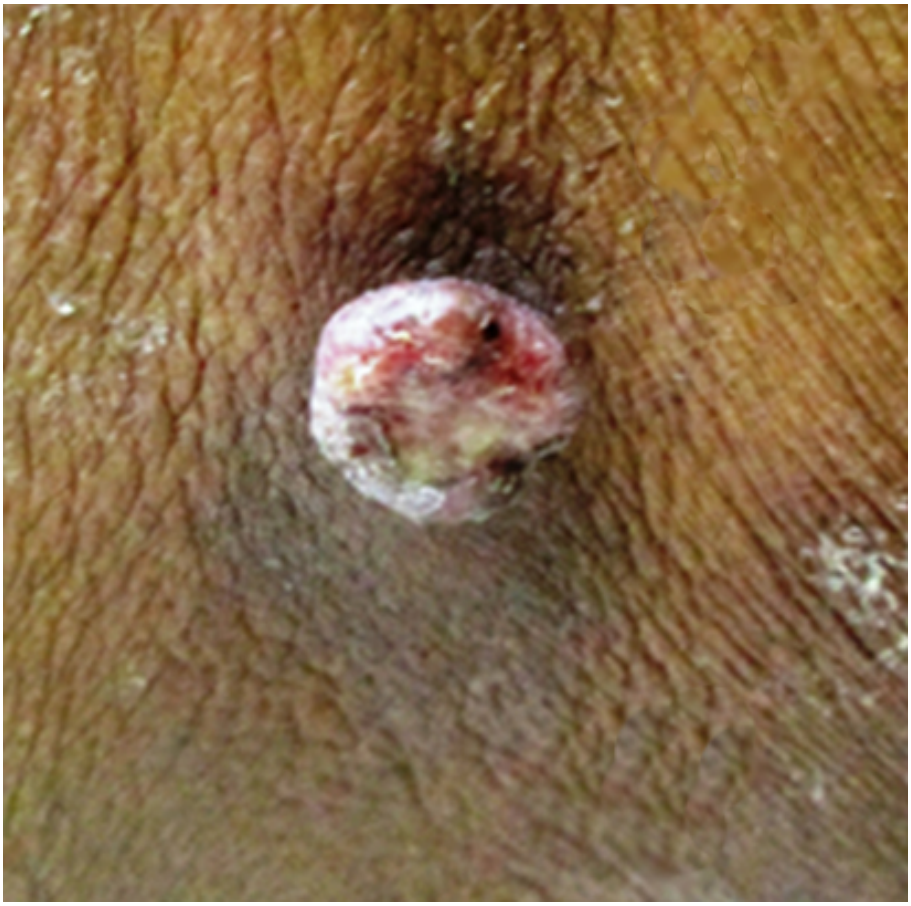
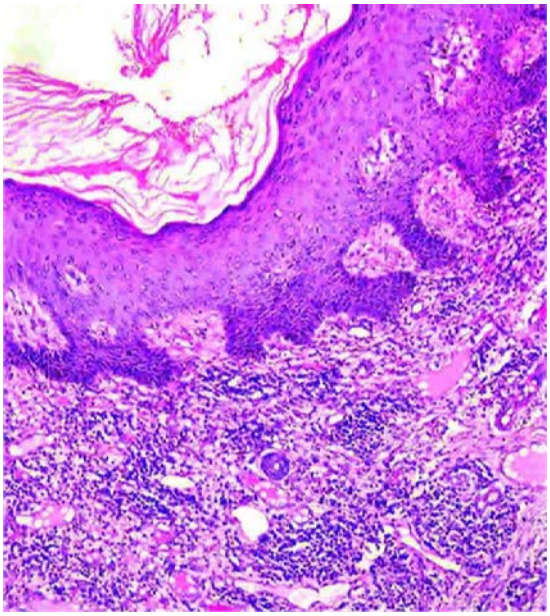


Figure 1. A single ulcerated nodule on the back with an inflammatory rim.

Examination revealed a single skin colored to erythematous ulcerated nodule located just left

to the midline on the lower back, approximately five centimeters(cm) in diameter. The surface showed evidence of hemorrhage at a few areas and there was a rim of erythema surrounding the base (Figure 1). On palpation, the nodule was non-tender, the erythema was not blanchable and the base was not indurated.

Differential diagnoses of keratoacanthoma, pyogenic granuloma, and basal cell carcinoma were entertained and an excisional biopsy was performed. Haematoxylin and eosin staining of the biopsy revealed a dermal vascular proliferation with abundant plasma cells infiltrating the stroma (Figures 2, 3). The vessels were abnormally large and the endothelial cells appeared plump, with an epitheloid appearance at a few places. Additionally the plasma cells showed no evidence of atypia or mitotic activity and stained positively in equal proportions for both kappa and lambda chains (Figure 4 and Figure 5).



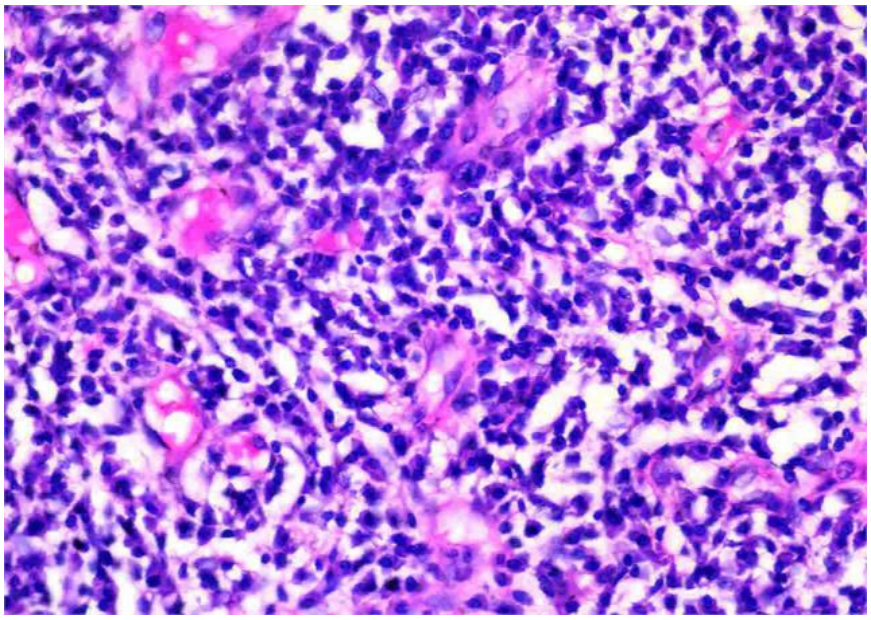
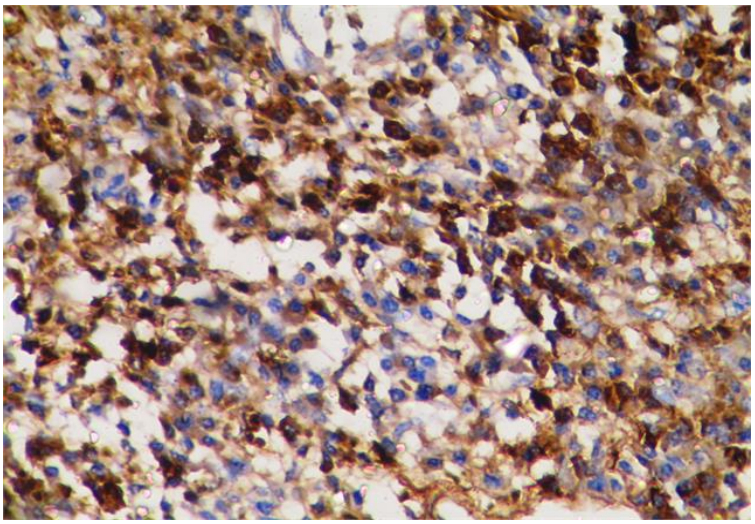


Figure 2. Haematoxylin and eosin staining (100 X) showing the dilated dermal vasculature with numerous plasma cells. **Figure 3.** Haematoxylin and eosin staining (400 X) showing prominent inflammatory infiltrate composed of plasma cells without atypia or increased mitoses



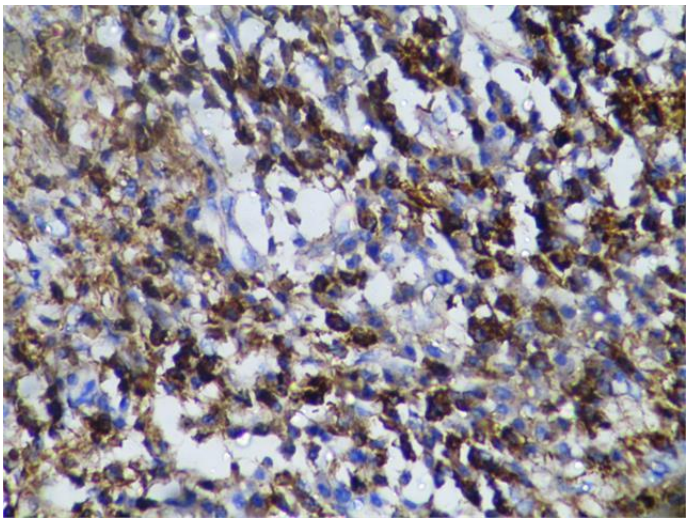


Figure 4. Immunohistochemistry: Kappa staining of the plasma cells (400 X). **Figure 5.** Immunohistochemistry: Lambda staining of the plasma cells (400 X)

Tissue culture, Periodic acid-Schiff, Giemsa, and Warthin-Starry stains were negative for any organism. Screening for myeloma including serum and urine electrophoresis revealed no abnormality. Routine investigations and chest X-ray revealed no abnormality.

The clinical features of the nodule along with its histopathology showing polyclonal staining of plasma cells without any evidence of atypia and the absence of any evidence of monoclonal gammopathy led us to the final diagnosis of angioplasmocellular hyperplasia. Seven months after excision there was no recurrence.

Discussion

Plasma cells are known to be present in normal mucous membranes and reactive plasma cell proliferation involving the mucosa has been well described [3]. By contrast, plasma cells are rarely found in normal skin. Purely cutaneous plasmacytosis without mucosal involvement is rare [4].

The majority of the primary cutaneous angioplasmocellular hyperplasia cases reported in the literature –have been isolated cases [5, 6] with the exception of the series published by Hsiao et al [2]. Hsiao and Wu in their series of 10 cases have expanded on what is known both clinically and histologically about angioplasmocellular hyperplasia. They believed that the clinical setting and histopathology is sufficiently distinctive to allow a definitive diagnosis of this clinical entity. A relationship with previous injury or with varicella has been hypothesized in few cases and it appears to be a form of reactive inflammatory vascular hyperplasia, although it is still not possible to define a definite inciting stimulus [2].

The condition is clinically characterized by erythematous-violaceous or brownish papules or nodules measuring between a few millimetres to few centimetres in diameter, occurring most commonly on the head and trunk. The lesions are usually solitary, although one reported patient had 2 simultaneous lesions [2]. Clinically central ulceration with a peripheral rim of erythema is a common finding. The diagnosis is usually delayed because of the asymptomatic nature of the condition [1, 2, 6].

The characteristic histopathology findings are dilatation and proliferation of dermal vessels with an inflammatory infiltrate consisting of predominantly polyclonal plasma cells (>60%) along with a small proportion of lymphocytes, neutrophils, and eosinophils without atypia [7].

No specific treatment modalities have been defined. However, the majority of cases reported have been managed by excision and no recurrence or systemic involvement was noted on follow up [1, 2].

Conclusion

To the best of our knowledge fewer than 20 cases of cutaneous angioplasmocellular hyperplasia have been published in the literature. Thus, we report this case of angioplasmocellular hyperplasia owing to its rarity. We would like to emphasize the importance of clinico-pathological correlation to differentiate it from various other conditions of cutaneous plasma cell infiltration.

References

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