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Case Report

Serpentine supravenous hyperpigmentation – A rare cutaneous manifestation in non-neoplastic conditions

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ABSTRACT

Serpentine erythematous or hyperpigmented streaks along with the superficial venous network usually occur as a distinctive eruption after infusion of several chemotherapeutic agents. This morphological pattern has been described under various terms such as "persistent supravenous erythematous eruption," "persistent serpentine supravenous hyperpigmented eruption," or "persistent serpentine supravenous hyperpigmentation (SSH). We describe four patients with no history of malignancy or treatment for malignancy who developed SSH.

Keywords: Supra venous pigmentation, Serpiginous pigmentation, Non-neoplastic conditions

INTRODUCTION

Serpentine supravenous hyperpigmentation (SSH) was the term coined by Hrushesky in 1976 to describe increased pigmentation of the skin immediately overlying the venous network.[1] The other names by which it has been described are "persistent supravenous erythematous eruption,"[2] "persistent serpentine supravenous hyperpigmented eruption," and "persistent SSH." It is a benign and self-limiting condition. Typically described as an eruption after infusion of various chemotherapeutic agents, SSH was later rarely reported in leprosy,[3] hemolytic anemia,[4] and collagen vascular diseases.[5] Around 18 cases have been reported worldwide to the best of our knowledge.[2,6-9] We are presenting four different non-neoplastic conditions with linear pigmentation along superficial leg veins.

CASE REPORTS

Case 1

Our first patient was a 65-year-old male who presented with recurrent episodes of itching and oozy lesions of legs which were of three years duration with exacerbations during the cold season. The lesions subsided with depigmentation. The present exacerbation was of 1-month duration. He gave a history of treatment with topical and oral medications for these lesions, and the current exacerbation was treated with topical and oral antibiotic and oral steroids from another hospital. The patient had no systemic comorbidities. On examination, there were well-ill-defined plaques showing erosions, crusting, oozing, and mild scaling in few areas. On the anterior aspect of the lower one-third of leg, there were multiple depigmented macules and linear hyperpigmented streaks along the superficial veins. The veins underlying were neither tender nor thickened.

Investigations showed a mild increase in total count and eosinophilia. Stool routine showed roundworm ova and strongyloidiasis. Screening for human immunodeficiency virus was negative. The clinical diagnosis was asteatotic eczema [Figures 1 and 2].

Case 2

The second patient was a 41-year-old female who presented with depigmented lesions on hands, legs, ankles, and feet. There was no history of eczema or any systemic comorbidities. The diagnosis was Vitiligo vulgaris [Figure 3]. She gave a history of use of topical psoralen for treatment of depigmentation but with no response. On examination, there were hyperpigmented linear streaks along superficial veins overlying the vitiliginous area. Her routine investigations were normal.

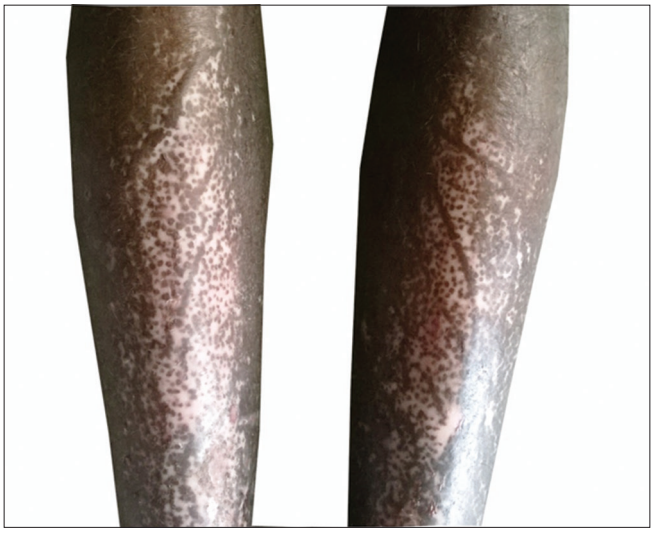

Figure 1: Pigmentation overlying the veins on both legs in a patient with eczema.

Figure 2: First patient with subacute eczema seen near the ankle.

Case 3

The third patient was a 43-year-old female who presented with induration of skin over face and limbs, non-healing ulcers on tips of fingers which were of 7 years duration, exertional dyspnea of 2 months duration and dysphagia to solid foods. She was hypertensive for the past 19 years. On examination, there was sclerosis of face, forearms, arms, trunk, and legs, salt and pepper pigmentation on neck, ears, chest abdomen, and lower back. Mask facies with telangiectasia on the face, sclerodactyly, and digital pitted scars were also present. On legs, there were areas of depigmentation with spotty pigmentation and linear hyperpigmented streaks along the superficial veins. Serology for antinuclear antibody and Scl 70 was positive. High resolution computed tomography of the chest showed early interstitial lung changes. Venous Doppler of the left lower limb showed evidence of chronic thrombosis with recanalization. She was diagnosed as diffuse systemic sclerosis [Figures 4 and 5].

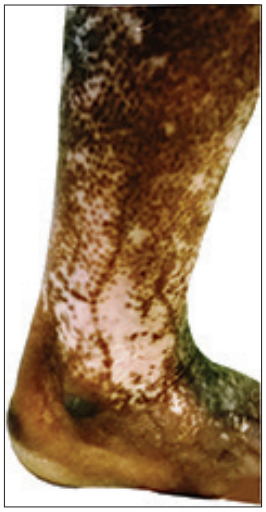

Figure 3: Pigmentation overlying the veins in the patient with vitiligo.

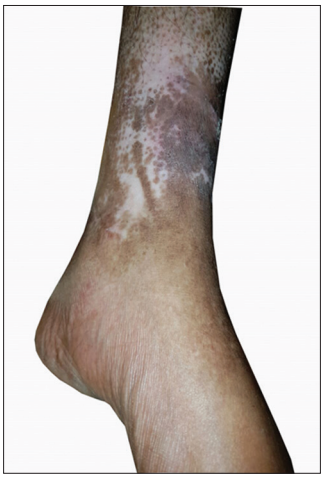

Figure 4: Pigmentation overlying veins in a patient with systemic sclerosis, sclerosis of the skin can be noticed.

Case 4

The fourth patient was a 50-year-old female who presented with pruritic hyperpigmented and erythematous plaques seen extensively on the trunk and lower limbs. She was treated from elsewhere with systemic terbinafine, griseofulvin, and other antifungals. Scraping and KOH examination from lesions revealed fungal hyphae. She was diagnosed as having extensive dermatophytosis [Figure 6]. On examination, in addition to the annular plaques, there were multiple liner streaks along the superficial veins of legs.

DISCUSSION

SSH appear either as hyperpigmented linear streaks preceded by erythema or as hyperpigmented streaks without erythema. They follow the superficial venous network in a serpentine or linear pattern. Usually, it is seen after intravenous use of chemotherapeutic agents such as 5-fluorouracil, docetaxel, cytosine arabinoside, daunorubicin, 6-mercaptopurine, cyclophosphamide, doxorubicin, bleomycin, vinca alkaloids, and dacarbazine.[2,6-9] One report suggests that it can either be a normal racial variant or associated with HIV.[10]

The precise mechanism for the pigmentation is not known. The probable cause for the pigmentation is (a) the extravasation of the cytotoxic agent following endothelial damage, causing epidermal basal hyperpigmentation, and dermal melanin incontinence.[1,6,11] Other hypotheses are (b) post-inflammatory hyperpigmentation of overlying skin following subclinical thrombophlebitis[11] or (c) due to the promotion of melanin synthesis through removal of inhibitors of tyrosinase by certain drugs or direct stimulation of melanocytes.[3] SSH is also rarely reported in HIV, collagen vascular diseases, hemolytic anemia, and lepromatous leprosy. In leprosy, the pigmentation is attributed either to the post-inflammatory hyperpigmentation caused by phlebitis induced by the disease process or to drugs such as rifampicin, minocycline.[3] Our patients were diagnosed with eczema, vitiligo, scleroderma, and extensive dermatophytosis. The exact pathogenesis of the SSH in these dermatoses remains unknown. It might be a coincidental finding or a rare association which requires further research. Eczematous dermatoses, connective tissue diseases, and dermatophytosis have significant inflammatory responses (either acute response as in eczemas or chronic inflammation as in connective tissue diseases), following which there is the release of pro-inflammatory mediators resulting in vasodilatation, increased permeability, and activation of endothelial cells. The loss in endothelial integrity and mild phlebitis may probably result in melanin incontinence and basal hyperpigmentation. Jawitz *et al.*[12] had reported pigment retention over the superficial blood vessels in areas of depigmentation in three patients with systemic sclerosis and attributed it to a local thermal mechanism which might be a cause for the retention of supravenous pigment in vitiligo and scleroderma as observed by us.

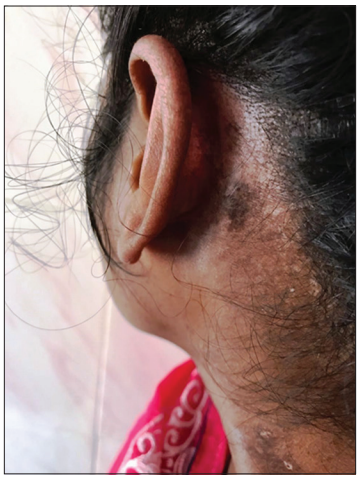

Figure 5: Salt and pepper pigmentation on the scalp in a patient with systemic sclerosis.

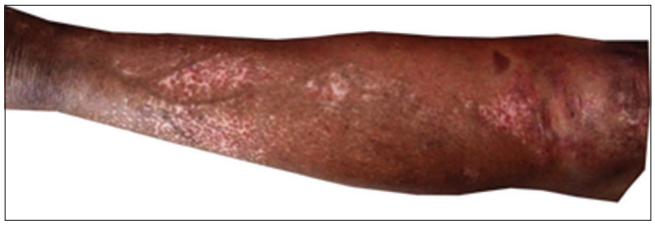

Figure 6: Pigmentation overlying veins in a patient with dermatophytosis. Dermatophytic plaque on the knee.

CONCLUSION

The main aim of reporting these cases is to increase the awareness regarding supravenous pigmentation. The appearance of SSH in the above-mentioned dermatological conditions is hitherto unreported or very rarely reported as in scleroderma. Dermatologists may perhaps keep a keen eye for the same.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s)/have given their consent for 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.

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Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Hrushesky WJ. Serpentine supravenous fluorouracil hyperpigmentation. JAMA 1976;236:138.
2. Pujol RM, Rocamora V, Lopez-Pousa A, Taberner R, Alomar A. Persistent supravenous erythematous eruption: A rare local complication of intravenous 5-fluorouracil therapy. J Am Acad Dermatol 1998;39:839-42.
3. Narang T, Dogra S, Sakia UN. Persistent serpentine supravenous hyperpigmented eruption in lepromatous leprosy after minocycline. Lepr Rev 2015;86:191-4.
4. Avarbock AB, Gill KZ, Lauren CT, Chong DH, Silvers DN, Grossman ME, *et al.* Serpentine supravenous hyperpigmentation secondary to superficial venous thrombosis in autoimmune hemolytic anemia. Int J Dermatol 2014;53:e96-7.
5. Werth VP, Schumacher HR Jr., Von Feldt JM. Linear hyperpigmentation overlying veins in association with collagen vascular disease. J Am Acad Dermatol 1993;29:1039-40.
6. Geddes ER, Cohen PR. Antineoplastic agent-associated serpentine supravenous hyperpigmentation: Superficial venous system hyperpigmentation following intravenous chemotherapy. South Med J 2010;103:231-5.
7. Ghosh SK, Bandyopadhyay D, Ghoshal L, Basu S. Letter: Docetaxel-induced supravenous serpentine dermatitis. Dermatol Online J 2011;17:16.
8. Das A, Kumar D, Mohanty S, Mondal AK, Chowdhury SN, Bandyopadhyay D, *et al.* Serpentine supravenous hyperpigmentation induced by docetaxel. Indian J Dermatol Venereol Leprol 2015;81:434.
9. Chan CC, Lin SJ. Images in clinical medicine. Serpentine supravenous hyperpigmentation. N Engl J Med 2010;363:e8.
10. O'Malley JT, Lieb JL, Weiser JA, Grossman ME. Persistent serpentine supravenous hyperpigmentation a possible cutaneous manifestation of HIV infection or a normal racial variant: A report of 3 cases. Skinmed 2013;11:93-4.
11. Aydogan I, Kavak A, Parlak AH, Alper M, Annakkaya AN, Erbas M, *et al.* Persistent serpentine supravenous hyperpigmented eruption associated with docetaxel. J Eur Acad Dermatol Venereol 2005;19:345-7.
12. Jawitz JC, Albert MK, Nigra TP, Bunning RD. A new skin manifestation of progressive systemic sclerosis. J Am Acad Dermatol 1984;11:265-8.

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