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Hematidrosis

SUBHAM BHATTACHARYA, MRINAL KANTI DAS, SUMAN SARKAR, AND *AVISHEK DE
*From Department of Pediatric Medicine and *Dermatology, IPGMER & SSKM Hospital, Kolkata, West Bengal, India.*

Correspondence to:
Dr Subham Bhattacharya, 39 Bhadreswar Station Road(S). Hooghly. 712124. P.O-Saradapally, India. drsb1979@yahoo.co.in
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Hematidrosis is an extremely rare clinical entity characterized by recurrent episodes of skin bleeding mixed with sweat. We report a case of hematidrosis in a 10-year-old girl where detailed laboratory and histopathological investigation revealed no abnormality. The girl was successfully treated with propranolol with no recurrence of bleeding over a follow-up of 3 months.

Keywords: *Child, Hematidrosis , Propranolol.*

Hematidrosis is a extremely rare and enigmatic disorder characterized by recurrent episodes of self limited bleeding from skin. Though, classically hematidrosis means blood in sweat, practically blood is mixed with sweat like material rather than true sweat in this condition [1]. Till date only nine cases are reported in literature. We report a case of hematidrosis diagnosed and treated successfully.

CASE REPORT

A 12-year-old girl was referred to our hospital with complaints of recurrent episodes of spontaneous skin bleeding for last one month. The bleeding was spontaneous from different sites of body including face, limb, palm and sole but not from mucous membrane. The consistency of bleeding was little thinner than blood and stopped as soon as the site was wiped, leaving behind no oozing site. It was occurring both day and night but never after she slept.

During hospitalization she had more than 10 instances of spontaneous intermittent bleeding per day that was evidenced by almost all of our on duty doctors and nurses. Her medical records were non-contributory with no known underlying disease. Her development was normal with normal tanners staging and menarche not yet started.

Complete blood count, and blood biochemistry was normal, with normal coagulation screening tests. Platelet aggregation test and estimation of von-Willebrand factor revealed no abnormality. Assay of anti-nuclear antibody and interleukins was done with a view to exclude underlying autoimmune conditions like vasculitis, but revealed no abnormality. Microscopic examination of bloody exudates from face revealed the same components as of peripheral blood including red blood cells, leukocytes and platelets mixed with epithelial cells. Skin biopsy performed from the bleeding area in palm immediately after the bleeding revealed irregular acanthosis of epidermis with broadening of rete pigs along with hypergranulosis and marked hyperkeratosis with edema of superficial dermis, similar description toin a previous case [3]. There was no abnormality in sweat gland or sebaceous gland.

The girl was treated initially with diazepam without any significant improvement. Finally, based on a previous case [5], she was treated with propranolol in a dose of 1 mg/kg/day in two divided dose with a baseline ECG and monitoring of heart rate. The bleeding episodes started to reduce within 2 days of therapy and stopped completely within 6 days. There was no recurrence of bleeding within three months of follow-up.

DISCUSSION

Hematidrosis is a condition in which capillary blood vessels that feed the sweat glands rupture, causing them to exude blood [2]. The term hematofolliculohidrosis has

been proposed because it appears along with sweat-like fluid and the blood exudes via the follicular canals [3]. Although, historically it was described to occur under conditions of extreme physical or emotional stress, various case report have shown that it may occur without stressful situations, as in our case. Recently, sympathetic nerve ending activation and vasculitis have been proposed as an important cause. The etiopahogenesis of this condition was described as bleeding from the blood vessels that supply the sweat gland [4]. Common differential diagnosis is self-injury and chromhidrosis (sweat containing color pigment). In our case there was no evidence of self-injury and diagnosis established with demonstration of blood corpuscles in the secretion along with negative tests for bleeding diathesis. Vasculitis was also excluded in this case with normal ANA and interleukin levels and finally normal skin biopsy.

Till date there is no specific management. Vitamin C and hemostatic drugs are not effective. Manolukul, *et al.* [2] used lorazepam as anxiolytic in a case and got excellent result. Zhaoyue, *et al.* [5] used propranolol with the hypothesis of sympathetic overactivity and it was found to be effective. In our case diazepam did not work, but we got excellent result with proapranolol. Bleeding stopped completely within one week of starting treatment and did not recur over next three months of follow up.

Contributors: SB, MKD, AC, SS, AD were involved in diagnosis and management of the case. SB drafted the paper and searched literature. SB, MKD & AD contributed to paper writing. All author approved the final manuscript.
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