Case Report

Exploring Hematohidrosis: Unveiling the Mysteries of a Rare Condition

Tejaswani Patel, Krishnendra Varma, Ujjwal Kumar, Shashank Bhargava*

Hematohidrosis is a rare condition characterized by the excretion of blood through eccrine sweat glands without associated injury or trauma. Despite various theories, its exact cause remains unclear. Diagnosing this condition can be challenging, as it requires ruling out bleeding disorders, vasculitis, and related conditions. Here, we present a case of hematohidrosis in a 14-year-old male who experiences multiple daily episodes without identifiable causes. The patient has undergone various treatments but has not yet achieved remission.

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INTRODUCTION

Hematohidrosis, or hematidrosis, is a rare phenomenon where individuals sweat blood from intact skin and mucous membranes, without any skin injury or trauma.[1] This disorder is extremely uncommon, with few documented cases in medical literature.

Many cases are linked to acute fear, chronic stress, or anxiety, though some healthy individuals develop it without clear triggers. Despite its alarming presentation, hematohidrosis is generally considered benign and often transient, resolving on its own over time. Due to the limited number of cases documented, further research is necessary to understand its underlying mechanisms and treatment options.

Case Report

A 14-year-old Indian male presented to our dermatology outpatient department with a history of unusual, painless bleeding from various body parts, including the face, eyes, arms, legs, and trunk, beginning in February 2023. These spontaneous, intermittent, and unpredictable episodes occurred without identifiable triggers and showed no correlation with mood, activity, or sleep patterns.

Each episode was preceded by an aura of pain and tingling sensations, lasting from seconds to several minutes, ceasing upon wiping, and leaving no residual scars. He also reported instances of epistaxis and hematemesis. The patient's medical history was otherwise unremarkable, with no trauma, drug abuse, allergies, hematological disorders, or systemic diseases. He denied taking anticoagulants or medications, and a thorough examination revealed no signs of trauma. Their family history was negative for bleeding disorders.

Upon evaluation, the patient was alert, oriented, and communicative, with no psychotic symptoms detected. His vital signs were normal, and he appeared healthy. Cutaneous examination showed a few ecchymotic patches on the left forearm, but no active bleeding. Laboratory investigations returned normal results, except for a positive benzidine test of oozed blood, indicating the presence of hemoglobin.

Initially, the patient was prescribed propranolol (20 mg once daily), vitamin C, vitamin K, alprazolam (0.25 mg), and glycopyrrolate (1 mg twice daily) for three months; however, these treatments were ineffective. Alternative medications also failed to provide relief.

Department of Dermatology, RD Gardi Medical College, Ujjain, Madhya Pradesh, India.

Correspondence to: Tejaswani Patel, Department of Dermatology, RD Gardi Medical College, Ujjain, Madhya Pradesh, India. E-mail: tejaswanipatel1106@gmail.com

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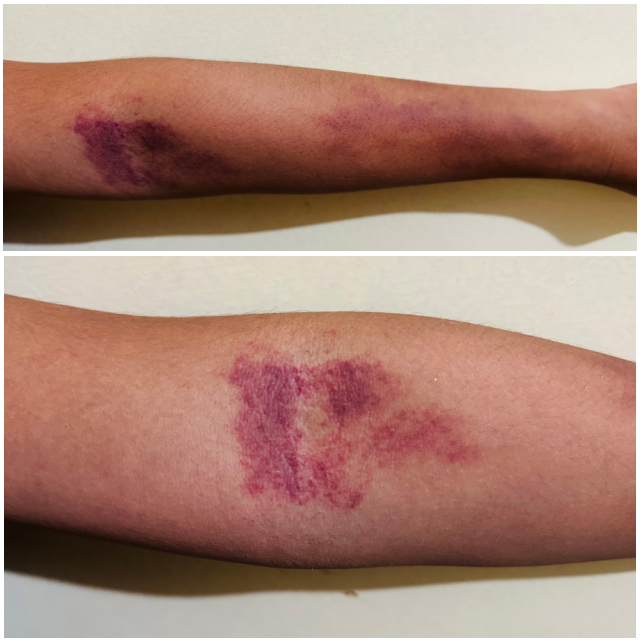

Fig. 1: Patient's left forearm at the time of examination

DISCUSSION

Hematohidrosis is a perplexing disorder characterized by recurrent episodes of bleeding from intact skin. It is documented in a limited number of cases, with some researchers proposing the term "hematofolliculohidrosis," as bleeding often occurs alongside sweat, exuding through hair follicles.[2]

Most reports indicate that hematohidrosis primarily affects females, accounting for approximately 84% of cases, with ages ranging from 9 to 72 years.[3] The median age at presentation is around 13 years, particularly among female pediatric patients. This case differs from typical demographics and aligns with the observation that most documented cases involve individuals of Asian descent.

Clinically, hematohidrosis manifests as oozing blood from the skin or mucous membranes, usually triggered by stress or anxiety.[2] Despite its distressing impact, the diagnosis is often delayed due to its rarity and the presence of multiple differential diagnoses. Patients frequently undergo extensive evaluations, including consultations with hematologists, before reaching a definitive diagnosis.

The etiopathogenesis of hematohidrosis remains an area of ongoing research, with no single explanation adequately accounting for the source of the bleeding. One prevailing theory suggests that stress-induced vasoconstriction triggered by sympathetic stimulation affects blood vessels around the sweat glands, leading to vascular rupture and subsequent blood extravasation

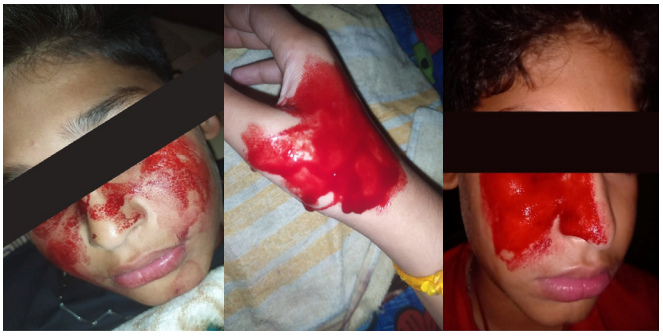

Fig. 2: Patient sweating blood from face and right hand

alongside sweat.[2] Some researchers, including Manonukul *et al*.[4] have suggested that dermal defects might create channels connecting to vascular spaces, allowing blood to accumulate and drain through follicular canals or directly onto the skin's surface.

Dr. Frederick Zugibe[5] proposed that a net-like arrangement of blood vessels surrounding sweat glands constricts under stress, and as anxiety subsides, these vessels may dilate excessively, leading to rupture and blood entering the glands, which is then pushed to the surface. The persistent pain and tenderness experienced by the patient after each bleeding episode may be linked to underlying vasoconstriction. Additionally, Zhang *et al*.[6] noted obstructed capillaries and intradermal bleeding, suggesting that vasculitis could be a contributing factor.

Postulated pathophysiology of hematidrosis

Hematidrosis is explained through several proposed mechanisms. One theory suggests that inflammation in the dermal blood vessels, triggered by intense stress and anxiety, leads to constriction and subsequent dilation of the vessels surrounding sweat glands. This may result in blood leaking into the sweat ducts. Another explanation

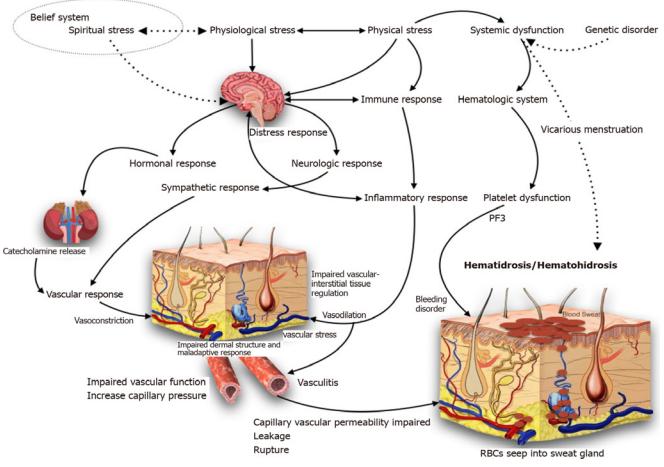

Figure 1: Pathophysiology of Hematohidrosis

indicates that the blood vessels near the sweat glands are organized in a network. Under extreme stress, these vessels can contract, and when the stress alleviates, they may dilate excessively, causing rupture. This rupture allows blood to enter the sweat glands, which then forces it to the surface, creating droplets of blood mixed with sweat.[7]

Diagnostic investigations typically return normal results, with an examination of the bloody fluid revealing blood components. Overall, the viability of a skin biopsy for diagnosing hematohidrosis remains unclear. A thorough skin examination is crucial to rule out self-inflicted injuries and differentiate hematohidrosis from conditions like chromhidrosis and vasculitis.

Currently, no specific therapy exists for hematohidrosis, although treatments such as anxiolytics have shown efficacy in stress-triggered cases. Several authors have noted positive responses to propranolol at a dose of 1-mg/kg/day, supporting the role of sympathetic nerve activity in the condition's pathogenesis. While many reported cases show bleeding episodes triggered by stress, no identifiable triggers were found in the current case. Recently, Biswas *et al*. reported a successful case treated with a transdermal atropine patch, indicating potential new avenues for management.

We present this case due to its rarity in a male child and the failure of standard treatments available in the literature to provide relief. This highlights the complexities of hematohidrosis and the need for innovative management approaches This case serves as a reminder of the limitations of current medical knowledge and the need for ongoing research into hematohidrosis.

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