Galore International Journal of Health Sciences and Research
Vol. 9; Issue: 1; Jan.-March 2024
Website: www.gijhsr.com
Case Report *P-ISSN: 2456-9321*

Hematohidrosis: Blood, Sweat and Stress

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DOI: https://doi.org/10.52403/gijhsr.20240120

ABSTRACT

Hematohidrosis or hematidrosis, a rare disorder, involves spontaneous episodes of blood-stained sweat/secretion from intact skin or mucous membranes, particularly from eccrine glands. Though its etiology is unclear, it is often believed to manifest under extreme stress. The condition predominantly affects young individuals, especially from Asian countries like India. This case report focuses on a 10-year-old boy from Tiruppur district, Tamilnadu in India with blood-stained sweat/secretion oozing from various sites of the body. Diagnostic evaluations ruled out bleeding disorders, auto-immune diseases, renal artery stenosis, and other systemic diseases. Hematohidrosis was confirmed based on blood investigations and clinical presentation. Underlying stress associated with past experiences in school was identified as a trigger factor. This study explores the efficacy of a multidisciplinary approach, including pharmacotherapy, psychotherapy, and medical nutrition therapy in managing this unusual condition. De-stressing counselling given to the child in addition to nutrition counselling reportedly had a positive impact in mitigating the stress of the child and subsequent improvement in hypertension. A follow-up observation of the patient for nearly one and a half months revealed no episodes until three weeks after discharge followed by negligible frequency of episodes. The patient is being followed up regularly for further management of this clinical condition. This report contributes to the understanding of hematohidrosis and its intervention, emphasizing the importance of multi-disciplinary team approach and reassurance to the patients and their families about the transient nature of this condition.

Keywords: hematohidrosis, hematidrosis, blood-stained sweat, eccrine gland, stress, nutrition counselling

INTRODUCTION

Hematohidrosis, also known as hematidrosis or hemidrosis is a rare and mysterious phenomenon that is characterized by one or more frequent episodes of spontaneous, blood-stained sweat/secretion from intact skin surfaces and/or mucous membranes, unaffected by trauma[1]. Historically, this condition was reported, notably in one of the descriptions written by Leonardo Da Vinci about a soldier who sweated blood on his way to the battle ground and also in Jesus Christ who sweated blood while praying under extreme stress before crucifixion [7]. Though, its exact systemic etiology remains elusive, however activation of the sympathetic nervous system owing to increased stress is found to be involved. Under extreme

conditions of stress and fear, the net-like form of tiny capillary blood vessels that surrounds the sweat glands constrict and as the stressor subsides, dilate to the point of rupture. This in turn, causes the blood to diffuse into the sweat glands, thereby presenting as droplets of blood mixed with sweat on the skin surfaces [1]. Under the International Classification of Disease 10 (ICD 10) in 2016, hematohidrosis/ hematidrosis is given a diagnosis code of L74.8 that comes under the class of "other eccrine sweat disorders". The head region is the most commonly site to be affected, and some patients may experience prodromal symptoms (early signs/symptoms that precede the actual episode) such as headache, giddiness and abdominal pain. However, patients may also present with gastrointestinal (GI) bleeding (hematemesis, hematuria, melena, hematochezia, or rectal bleeding), or epistaxis. Most blood-sweating episodes subside within a few seconds to a few minutes, although in some cases, the episode persists upto 30 minutes [3,12]. Symptoms, such as prodromal sensations or mucosal bleeding, along with identified triggers are often linked to stress factors within familial or social contexts. The diagnosis is primarily by exclusion after all the other diagnostic tests are normal. Other differential diagnoses, such as chromhidrosis, bleeding disorder, or self-inflicted skin lesions are considered while identifying this condition. In the cases reported so far, the progression was predominantly favorable, ranging from mild improvement to complete remission. Management with psychotherapy involving anxiolytic drugs, or pharmacotherapy with beta blockers has proven to be effective in most of the cases [2].

The number of reported cases of hematidrosis/ hemidrosis has substantially increased over the past 20 years. Moreover, it is striking that most of the cases are thoroughly concentrated in Asia, particularly India (*n* = 40/106) and China (*n* =11/ 106) [3]. It mainly affects young girls ranging between the ages of 9 and 15 [2]. Herein, we present a 10-year-old boy from the southern region of India with hematohidrosis and the efficacy of pharmacotherapy, psychotherapy, and medical nutrition therapy as part of the management of this rare disorder.

CASE PRESENTATION

A 10-year-old boy (height, 154.5 cm; weight, 54.0 kg) from Tiruppur district, Tamil Nadu, South India, presented to the Paediatric Out Patient Department in a Medical College Hospital with complaints of blood-stained sweat/secretion from various sites of the body for the past four months. The very first episode of blood-stained secretion was observed at the right thumb followed by other sites of the body.

Based on the caregiver's report, the dorsal wrist ***(Fig.1)***, external ear canal ***(Fig.2)***, eyelids, big toes, thumb fingers, elbows, knees, eyebrows, forehead ***(Fig.4),*** and oral cavity ***(Fig.3)*** were identified as sources of blood sweat/secretion.

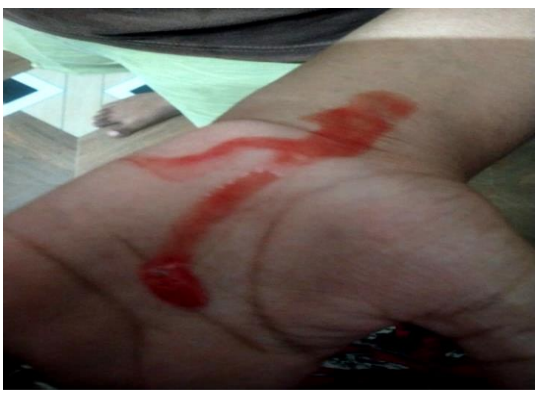

Fig 1. **Blood-stained sweat on the dorsal wrist**

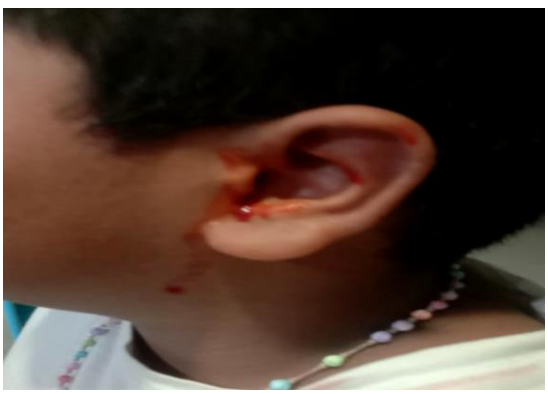

Fig 2. Blood stained discharge from the left auditory canal

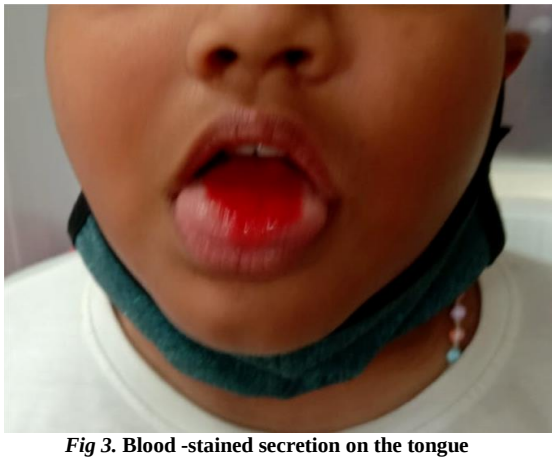

Fig 3. **Blood -stained secretion on the tongue**

Fig 4. **Blood-stained sweat on the forehead**

Frequent episodes of epistaxis and hematuria were also reported. The consistency of the discharge was sauce-like and sometimes watery, while the colour was deep orange/reddish orange except for the discharge from eyelids which was red. The episodes occur every week for at least three consecutive days and the frequency of episodes ranges from 4-5 times per day to a maximum of 20 episodes per day (occasionally). Each episode usually lasted from a few seconds to less than 2 minutes daily. Upon further investigation, it was found that prodromal symptoms (pricking or tingling sensation followed by chillness over the site prior to the episode) were strongly felt and described by the patient as a forewarning.

There were no complaints of pain over the site of discharge on the skin. Swelling of fingers and toes that resolved upon rest was reported. The patient also had complaints of giddiness, headache, and abdominal pain occasionally. Stress and irritability substantially raised the blood pressure of the child and were found to be possible triggers of the blood stained sweating episodes. The patient was hospitalized for further investigation and underwent a series of diagnostic tests to confirm hematohidrosis. The patient was subjected to evaluation for hematohidrosis, Von Willebrand Disease, and platelet function disorders. The results of the laboratory tests revealed a normal blood picture with occasional schistocytes, Prothrombin Time (12.7 s), Activated Partial Thromboplastin Time (32.4 s), International Normalized Time (0.93 s), and normal bleeding and coagulation time. Factor XIII activity assay was also performed. The results were within limits, thus Factor XIII deficiency disorder was excluded. ANA (Anti-nuclear Antibody) and ELISA (Enzyme Linked Immuno Sorbent Assay) test was found to be negative, ruling out the possibility of autoimmune disorders. Microscopic examination of the blood stained tear was found to be weakly positive. Consequently, the patient was diagnosed with hematohidrosis. Since hematohidrosis is triggered by emotional or physical stress, the patient was referred to a pediatric psychiatrist to rule out depressive disorders. The child acknowledged certain undesirable traumatic experiences (physical and mental abuse) at school, involving rebuking, beating, and similar punishments from his class teacher, despite excellent scholastic performances. The child had also experienced an episode of frank bleeding on the head region when hit by the headmaster of the school. Physical assault by the school authorities was an important trigger source of stress for him. A year after the incident, the child began to manifest blood-sweating episodes. Despite quitting school, the child remained stressed and worried about the state of his fellow students in the same school, all of which added to his mental stress and led to a subsequent rise in blood pressure and manifestation of blood sweating episodes. The child also had complaints of sleep disturbances. Both the

patient and his parents were counselled about the nature of this rare disorder and were advised to create a stress-free environment for the child.

Pharmacotherapy: Anxiolytic drug – T. Alprazolam 0.25mg was given for 2 days and called off. T. Clonazepam 0.5 mg was given for 4 days, called off in between, and then continued for 12 days until discharge. T. Nifedipine 20 mg TDS was started on Day 3 to manage essential hypertension and continued throughout the course of treatment. The patient was advised to continue the same dosage on discharge. T. Prazosin 2.4 mg TDS was started during hospitalization and the dosage was increased to 3.2 mg TDS at the time of discharge which further reduced to 1.5 mg TDS during follow-up (post renal artery angiogram). T. Enalapril 2.5 mg and T. Propranolol 40 mg were given for 14 days and 9 days respectively and then called off. Chlorpheniramine 4mg, T. Montek LC 10mg, T. Amoxyclav 625 mg, and cough syrup 5ml were administered to treat prolonged cold and wet cough and then called off once the symptoms subsided.

The number of episodes reduced from 4-5 episodes per day in the past 4 months before hospitalization to about 3 episodes in three weeks during the stay in the hospital. Nil episodes were recorded until 3 weeks post discharge after which there was a recurrence of one episode in the oral cavity. Two weeks later, the patient was readmitted to perform a **Digital Subtraction Angiograhy** (Renal) to rule out Renal Artery Stenosis.

The findings of the test revealed no significant stenosis noted in the bilateral renal artery. The patient was discharged and advised to continue T. Nifedipine 20 mg TDS and the dosage of T. Prazosin was reduced to 1.5 mg TDS. One episode of epistaxis and blood stained secretion on the tongue was reported again post-discharge. The patient is being followed up regularly to record the improvements. After 3 weeks of discharge T. Nifedipine 20 mg was reduced to BD instead of TDS.

Nutrition intervention: The patient was referred to the Clinical Nutrition Out Patient Department regarding diet modification. Before hospitalization, the patient was consuming approximately 1568 Kcal of energy and 46 g of protein per day. Dietary assessment evidenced poor dietary habits, involving frequent consumption of processed foods, excessive intake of pickles, and inadequate intake of fruits and vegetables. In addition to that, Class I Obesity (BMI-22.9 Kg/m^2), Impaired Fasting Glucose (FBG- 118mg/dl and 102mg/dl as of 26/11/2023 and 09/01/2024 respectively), poor blood pressure control and stress-eating were found to be problems of concern. Despite the blood loss during every blood sweating episode, the patient was not found to be anemic. To improve the metabolic health of the patient, the type of carbohydrate and fat was revised. A low glycemic index, low glycemic load, fibre rich, low saturated fatty acid, nil trans fat, glycemic-friendly, moderate sodium restricted, weight optimization diet was suggested in view of class I obesity.

Since the present total energy intake of the child is lesser than his total energy requirement, he was advised to maintain the present calorie intake, ensuring a balanced proportion of all macronutrients. Hence a diet providing 1500 Kcals, 49g of protein, and 21g of fibre per day was suggested. A reduction in quantity of carbohydrate and fat intake with well-balanced nutrient sources along with the elimination of carbonated beverages rich in simple carbohydrates was suggested. Further, reduced intake of processed, pre-packaged, and convenience foods high in saturated fat, salt and sugar was also emphasized. The patient was also encouraged to participate in at least 60 minutes of moderate physical activity daily, if not contraindicated. Given Stage 2 Hypertension (BP >95th percentile), sodium was restricted to <2300 mg/d. Further, healthy parenting practices associated with diet by promoting parental modeling of healthy eating behaviours were also emphasized [13,14,15].

Besides nutrition counselling, de-stressing counselling in the form of a pep talk addressing the stress of the child brought about a positive change in the child's behaviour, and a drastic reduction in the frequency and severity of the episodes with improvement in the blood pressure. Patient's caregiver reported marked improvement in the blood pressure as well as the psychological status of the child post nutrition and de-stressing counselling. An improvement in the blood pressure post discharge (**120/80, 109/56, 120/80** mmHg as of 17/12/23, 20/12/23 and 10/01/24 respectively) was noted unlike the values in hospitalization which at times reached as high as 180/90 mmHg. Blood pressure values tracked during the hospital stay and post discharge is provided in ***Fig.5.***

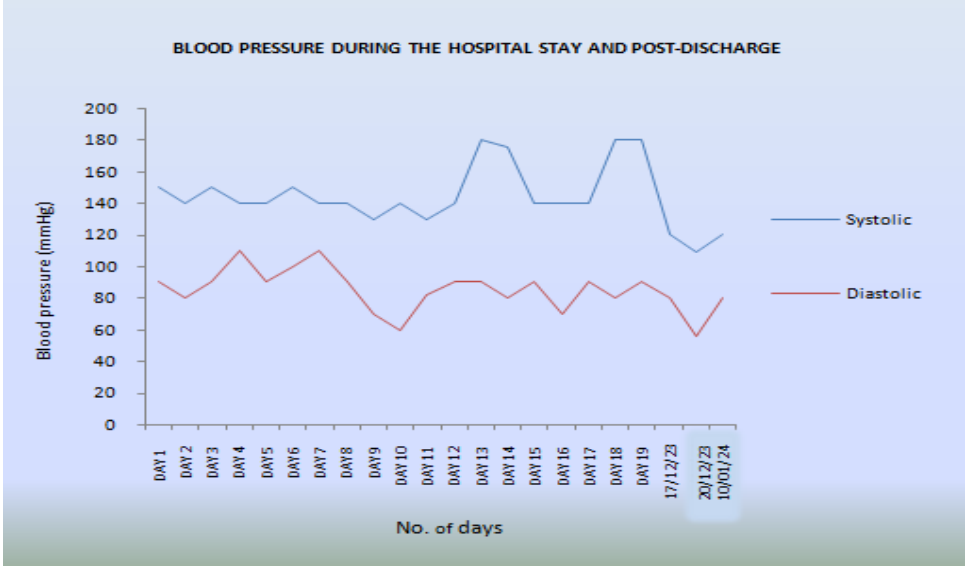

Fig.5 **Graphical representation of blood pressure tracked during the hospital stay and post-discharge**

Post Digital Subtraction Angiography (Renal), the patient was advised to follow a moderate calorie, sodium and fat-restricted, low protein diet @0.8g of protein/ kg present body weight with plenty of fluids for six weeks on account of the contrast dye used for imaging of the blood vessels to prevent contrast-induced Acute Kidney Injury [16]. Patient's adherence to healthy dietary practices is being monitored and documented during regular follow-up.

DISCUSSION

Hematohidrosis, or hematidrosis is an exceptionally rare condition where there is spontaneous blood-stained sweat/secretions from intact skin surfaces or mucosa across various body sites. Despite its extreme rarity and the lack of clear scientific evidence supporting its existence, diagnosis depends on the following criteria: (i) spontaneous, recurrent, painless, and self-limited oozing of blood sweat/secretion witnessed and confirmed by healthcare professionals, (ii) identification of typical blood components through laboratory investigations, and (iii) an intact bleeding site without abrasions, telangiectasia, or purpura, with no residual oozing after wiping the area. These criteria are crucial to exclude self-inflicted injuries, chromhidrosis (coloured sweat), bleeding disorders and factitious disorders.

In our case, no evidence of self-injury was found, and the diagnosis was confirmed by the presence of blood components in the secretion, coupled with negative tests for Factor XIII deficiency disorder and auto-immune disorders. The etiology and pathogenesis of hematohidrosis remain elusive. While fear and anxiety are commonly associated with this condition, it can also occur without apparent preceding stressful situations. Stressors encompass physical exertion, changes in environmental

temperature, emotional excitement, infant irritability, bullying at school, witnessing traumatic events, work and academic stress, parental discipline, and stress within family or personal relationships [1,2,3,9,10]. Increased anxiety leads to the rupture of blood vessels, allowing blood to diffuse into sweat glands, manifesting as droplets of blood mixed with sweat. In the presented case, no underlying systemic disease was identified, and episodes of blood-stained fluid discharge were associated with periods of anxiety, irritability, and stress in the child. Treatment for hematohidrosis remains challenging, commonly involving a combination of pharmacologic (beta-blockers, anxiolytics) and non-pharmacologic (psychotherapy, nutrition intervention, counseling and physical activity) approaches.

Of all the reported cases, only 41.1% have completely resolved from blood-sweating episodes. Spontaneous improvements can occur, and some patients respond to medications

like propranolol, anti-depressants, atropine transdermal patches, and gauze wipes with adrenaline. Remission often involves pharmacotherapy, supportive psychotherapy, medical nutrition therapy, family education, and improved child-rearing practices. In this case, medical treatment with alpha, beta-blockers, ACE (Angiotensin Converting Enzyme) inhibitors, antifibrinolytics, and antacids was proposed. Since hematohidrosis is often considered a psychosomatic disorder, reducing stress may help break the cycle of stress-induced physiological responses and decrease the likelihood of experiencing blood-stained sweating episodes. In this case, continuous assault on the child by authorities of the school was a source of mental stress to the child which presented as spontaneous sweating of blood from various sites of the body. Sound nutrition and de-stressing counselling effectively improved elevated blood pressure, elevated fasting blood glucose and blood sweating episodes reduced tremendously. This emphasizes the importance of holistic approach for the management of this clinical condition.

CONCLUSION

In conclusion, hematidrosis, though its pathophysiology remains unclear, is often viewed as a somatization disorder predominantly affecting young children in developing countries. Its potential impact on patient's lives necessitates careful consideration and diagnosis to differentiate it from other medical conditions. While it can be a distressing experience for the patient and his/her family, raising awareness and reporting such cases can aid in global physician education, leading to improved management. Early detection, accurate diagnosis, and appropriate treatment, involving medical examination, analysis, and suitable interventions, including beta-blockers, anxiolytics, psychological support, medical nutrition therapy, and family support are crucial for favorable outcomes and complete remission.

Educating the patient and his/her family about the temporary nature of this condition plays a pivotal role in addressing and managing this enigmatic phenomenon. The key to successful treatment involves convincing the caregiver about the nature of this illness, the aggravating factors, and the viable treatment options. Understanding the connection between stress and the condition allows individuals to take proactive steps in avoiding or mitigating stressful situations. Given the peculiar nature of the disorder, a multidisciplinary approach plays a vital role in managing this condition and associated co-morbidities. To conclude, hematohidrosis is a benign, self-limited condition, but psychologically alarming for parents and patients. Physicians rarely encounter this condition, and thus, this report may reiterate awareness of the existence of a condition such as hematohidrosis and explains potential treatment strategies for effective management of this rare medical phenomenon.

Declaration by Authors

Conflict of Interest: The authors declare no conflict of interest.

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How to cite this article: Sachdev Meenakshi, Viveka, M, Lavanya, K. Hematohidrosis: Blood, Sweat and Stress. *Gal Int J Health Sci Res*. 2024; 9(1): 181-187. *DOI: https://doi.org/10.52403/gijhsr.20240120*

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