BANGLADESH J CHILD HEALTH 2021; VOL 45 (1) : 45-47

Haematohidrosis: A Rare Case Report of A 12 year Old Girl with Unexplained & Puzzling Spontaneous Multiple Site Bleeding

MAHBUB MUTANABBI[1], MD. ASIF ALI[2], SHOHELA AKHTER[3], TASKINA MOSLEH[4], JESMEEN MORSHED[5]

Abstract:

Hematohidrosis (bloody sweat) is a very rare clinical entity characterized by recurrent, spontaneous, self-limited episodes of blood oozing from intact skin or mucosa. A 12-year-old girl presented to our hospital with a history of repeated episodes of oozing of blood from different parts of the body like eyes, ear lobules, nose & navel. She also had history of episodes of hematemesis and haematuria. During examination, bleeding disappeared as soon as it was mopped leaving behind no sign of trauma. Most of these bleeding episodes occurred after stress. Her laboratory evaluation showed; bleeding time, clotting time and prothrombin time were normal. On the basis of clinical presentation and normal investigation, she was diagnosed as a case of hematohidrosis. We treated her with non-specific beta-blocker (propranolol). In spite of the rarity of hematohidrosis, it should be considered as a differential diagnosis of bleeding episodes in patients with normal physical and laboratory investigations.

Key words: *Haematohidrosis, Bleeding*

Introduction

Hematohidrosis (bloody sweat), also known as hematidrosis, is a rare disorder of unknown etiology, with only a few cases reported till date. It is characterized by recurrent episodes of self-limited exudation of bloody secretion from intact skin and mucosa. Some factors like somatoform and psychosomatic disorders are believed to induce bleeding, particularly acute physical or emotional stress, acute fear, anxiety and intense mental contemplation. Normal physical finding and laboratory investigations make this phenomenon difficult to understand fully.[1] Here we present a case of this rare condition and discuss possible mechanisms to explain their clinical manifestation.

Case Report

A 12-year-old girl studying in 7th grade was admitted to BSMMU hospital, Dhaka, Bangladesh on December 2019. She gave history of repeated bouts of hematemesis, hematuria along with episodes of oozing of blood from navel, eyes, ear lobules, and nose for last 4 months. It occurred in episodes, once or twice a week, sometimes more frequently. Each episode started with bubbly, viscous bright-red colored bloody secretion from above mentioned sites, lasted for about 3 -5 minutes and ceased spontaneously. On mopping, it disappeared immediately leaving no signs of trauma or any abnormal skin condition. Most of these episodes were preceded by a brief period of abdominal pain, vomiting or a headache. Due to this, parents stopped sending her to school and she was taken to various doctors.

She had no history of prolonged bleeding after cut injury, any skin rash, oral ulcer, photosensitivity, alopecia. She has not taken colored food or drinks, anticoagulants or anti- tubercular drugs or did not get any blood transfusion. She also did not have any history of using any topical medication or exposure to dyes. The pictures of pinkish staining of clothing, bedsheets, blood vomitus & blood oozing from eyes, ears, nose & umbilicus were taken during home and hospital stay. She was alert, conscious, oriented. Pallor, jaundice, bony tenderness, lymphadenopathy all were absent, anthropometrically she was well thriving. Skin survey was normal except occasional oozing of blood without any sign of injury, petechiae,

1. Professor, Department of Paediatrics, BSMMU, Dhaka.
2. Resident, Department of Paediatrics, BSMMU, Dhaka.
3. Associate Professor, Department of Paediatrics, BSMMU, Dhaka.
4. Assistant Professor, Department of Paediatrics, BSMMU, Dhaka.
5. Medical Officer, Department of Paediatrics, BSMMU, Dhaka.

Correspondence: Dr. Md. Asif Ali, Resident, Department of Paediatrics, Bangabandhu Sheikh Mujib Medical University (BSMMU), Bangladesh.Telephone no: +8801727211077, E-mail: asifali230789@gmail.com

Received: 22/09/2020 **Accepted:**15/02/2021

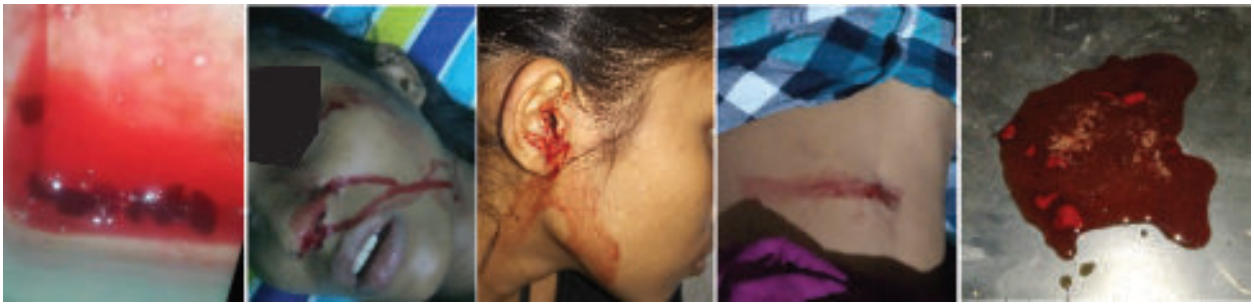

Fig-1: *Showing blood stained oozing from multiple sites: nose, eyes, ears, navel & also blood stained vomitus & clothing.*

purpura, ecchymosis or livedo reticularis. Vitally she was stable and systemic examination did not reveal any abnormalities.

At BSMMU hospital lab test results like CBC with PBF, prothrombin time, activated partial thromboplastin time, international normalization ratio, fibrinogen, Von Willebrand Factor assay, liver function test, kidney function test, random blood sugar, BT/CT were normal. Cytological test from bloody secretion revealed plenty of RBC along with 800/mm3 WBC which testifies the presence of blood in the secretion. Imaging studies like chest X-ray, endoscopy/colonoscopy, urine examination were done and found to be normal.

It was also found that she was temperamentally unstable. The episodes of bleeding from various parts of the body were preceded by issues such as upcoming exams, difference of opinion with parents, and parents not satisfying her demands. When she presented to us in December 2019, she was not going to school for the last 4 months due to the above symptoms.

The main focus of our treatment was nonpharmacological management that consisted of behavioral interventions for the child and counseling and psychoeducation to the parents, as it was clear that the stress precipitated episodes of bleeding. Relaxation exercises were taught to the girl to reduce anxiety. Parents were provided information about the illness; their undue concerns about the seriousness of the condition were addressed. They were explained about the antecedents of the bleeding episodes and how they should respond to such episodes like positive reinforcement, use of time out and token economy through which desired behavior would be encouraged and undesired behavior could be ignored. The plan we adopted for management of this case combined pharmacotherapy and nonpharmacological methods of treatment. We started treatment with tablet clobazam 10 mg at bedtime as an anxiolytic, tablet propranolol 10 mg thrice a day to block excess sympathetic discharge during stress and vitamin C supplementation for aiding proper hydroxylation of collagen fibers, specially in blood vessels.

Discussion:

Hematohidrosis is a very rare condition in which a human sweats blood. Blood usually oozes from the forehead, nails, umbilicus, and other skin surfaces. In addition, oozing from mucocutaneous surfaces causing nose bleeds, blood stained tears, and vicarious menstruation are common. The episodes are usually self-limiting. In some condition, the secreted fluid is more dilute and appears to be blood tinged, while others may have darker bright red secretions resembling frank blood.[1] Although haematemesis and haematuria is not a common manifestation of hematohidrosis, but case report described by Deshpande M. et al. in 2014 and also by Shahgholi E in 2018 also reported these symptoms in hematohidrosis patients.[2,3]

Diagnosis of hematohidrosis can only be made if the following criteria are met: (i) recurrent, spontaneous, painless and self-limited oozing of bloody discharge confirmed by health professionals, (ii) the usual blood components are found on biochemical studies of the discharge, and (iii) the site of bleeding is intact with no abrasion, telangiectasia or purpura and after wiping the area, there is no evidence of injury. All of these criteria must be met in order to rule out organic bleeding disorders, self-inflicted bleeding, factitious disorder by proxy, and chromhidrosis (colored sweat).[4]

Proposed etio-pathogenesis is that multiple blood vessels, which are present in a net-like form around the sweat gland constrict under pressure of stress. As the anxiety increases, the blood vessels dilate to the point of rupture. The blood goes into the sweat glands, which push it along with sweat to the surface,

presenting as droplets of blood mixed with sweat. The extravasated blood has identical cell components as that of peripheral blood.

According to Frederick Zugibe, under the pressure of stress, multiple bloods vessels which present in a net-like form around the sweat glands constrict and as the anxiety passes out, vessels dilate to point of rupture that induces RBCs released from blood vessels, go into the sweat glands and push to the surface.[5] Subsequently, they will collapse and leave no scar. This phenomenon, which acts like a balloon, will wax and wane; thus explaining why the bleedings are intermittent and self-limiting. The bleeding is intermittent because the vascular spaces will disappear after exuding their content but then recurred after the blood flow is reestablished.[6]

Another perspective of Zhang concluded that a specific vaculitis might be the pathological basis for hematohidrosis due to the presence of some intradermal bleeding and obstructed capillaries without abnormality in sweat glands, hair follicles and sebaceous glands.[7] In other side, Manonukul suggested that there maybe some dermal defects or some distinctive substances damaged the vessels supplying sweat glands.[6] Other researchers believed that this phenomenon could be related to some chronic underlying disease, namely Hemochromatosis.[8] Lack of evidence led to the difficulty in determination the cause of this disease.

The severe mental anxiety activates the sympathetic nervous system to invoke the stress-fight or flight reaction to such a degree as to cause hemorrhage of the vessels supplying the sweat glands into the ducts of the sweat glands.[7] Aggravating factors known to-date include emotional stress and excitement.

Physical and laboratory investigations do not lead to any abnormal results, making it difficult to fully understand this phenomenon.[1] An immediate biopsy is important for definite diagnosis. Biopsy during symptomatic period reveal blood-filled vascular spaces, intradermal bleeding & abnormality in hair follicles and sebaceous or sweat glands, but biopsy during symptom- free period does not reveal any abnormality in hair follicles and sebaceous or sweat glands.[7]

This condition is very rare but reports of successful treatment with beta blockers with a significant reduction in the frequency of spontaneous blood oozing are described by Manonukul J. et al. in their case report.[6] In our case too, propranolol was found to be effective in controlling the bleeding episodes. The successful use of beta blockers supports the theory that the condition is induced by stress and anxiety yet this etiology is also not well established. The high prevalence of stress and anxiety in the modern era did not change the incidence of this extremely rare disease suggesting that other co-abnormality also play a key role in this disease.[6] Recently, Biswas et al. have reported successful treatment with topical atropine application.[9] Symptoms of oppositional defiance were targeted through nonpharmacological interventions as described above. The key to successful treatment also includes convincing the parents about the nature of this illness, its aggravating factors and the possible treatment of this condition.

Conclusion:

We report this case for its rarity and also for its clinical response to propranolol, diazepam and vitamin C. In our case, the precipitating factor namely stress was identified. No evidence of vasculitis or any dermal defects or abnormality in sweat glands was detected. Despite its rarity, hematohidrosis should be considered as a miscellaneous differential diagnosis of bleeding episodes in a patient with normal physical and laboratory investigations. Such awareness can also play an important role to ensure a non-judgmental and compassionate approach by physicians. This is an important issue because an overzealous rush to accuse parents of factitious disorder can have tragic consequences for a family that is seeking help for their child who suffers from this very rare disorder. Further studies need to search for etiology and risk factors of such condition to correctly address clinical management.

References:

1. Holoubek JE, Holoubek AB. Blood, sweat and fear: A classification of hematidrosis. J Med. 1996; 27: 115-33.
2. Deshpande M, Indla V, Kumar V, Redd IR. Child who presented with hematohidrosis (sweating blood) with oppositional defiant disorder. Indian J Psychiatry 2014; 56 :289-91.
3. Shahgholi E. A case series of hematohidrosis: A puzzling medical phenomenon. Turk J Pediatr. 2018; 60: 757-61.
4. Bhagwat PV, Tophakhane RS, Rathod RM, Shashikumar BM, Naidu V. Hematohidrosis. Indian J Dermatol Venereol Leprol. 2009; 75: 317-18.
5. Freddrick Z. Hematidrosis. 2019.Available at http://www.enwikipediaorg/wiki/hematidrosis.
6. Manonukul J, Wisuthsarewong W, Nitiyarom R, Vongirad A. Hematidrosis: A Pathologic Process or Stigmata. A Case Report With Comprehensive Histopathologic and Immunoperoxidase Studies. Am J Dermatopathol. 2008; 30:135-9
7. Zhang FK, Zheng YL, Liu JH, Chen HS, Liu SH, Xu MQ, et al. Clinical and laboratory study of a case of hematidrosis. Zhonghua Xue Ye Xue Za Zhi 2004; 25: 147-50.
8. Jerajani HR, Jaju B, Phiske MM, Lade N. Hematohidrosis – A rare clinical phenomenon. Indian J Dermatol. 2009; 54: 290-92.
9. Biswas S, Surana T, De A, Nag F. A curious case of sweating blood. Indian J Dermatol. 2013; 58: 478-80.