ARCHIVES of NIMH
The Official Journal of National Institute of Mental Health, Dhaka

Case report

Hematohidrosis: a mysterious and rare disorder

Anika Ferdous, Faijul Islam, Taiyeb Ibna Zahangir

Abstract

Hematohidrosis is a mysterious and rare disorder characterized by one or more attacks of spontaneous, bloody sweating from intact surfaces of skin and/or mucous membranes. The exact explanation of this condition is not so clear, but activation of the sympathetic nervous system has been suggested. This is a case report of a 13-year-old girl with recurrent bleeding from right nostril, right ear and right eye for 1.5 months. She was depressed and had an anxious trait before these episodes. She was referred to National Institute of Mental Health as a suspected case of factitious disorder. She was worried about the significance of her symptoms, thinking she might have developed some serious illness. There was complete remission of bleeding with pharmacotherapy and psychotherapy.

Declaration of interest: *None*

Copyright and usage: ©Archives of NIMH 2020

Keywords: Hematohidrosis; etiology of hematohidrosis; factitious disorder

Introduction

Hematohidrosis or hematidrisis is the name given to the clinical phenomenon in which an individual sweats blood. Upon exposure to extreme anxiety, multiple blood vessels that form capillaries around the sweat glands undergo constriction and then dilatation to the point of rupture. Capillaries are tiny blood vessels located throughout tissue. They carry essential nutrients to different parts of the body. Capillaries are also located around the sweat glands. In cases of severe fear or stress, these tiny blood vessels can burst and cause blood to exit the body through the sweat glands. Then the blood goes into the sweat glands, which push it along with sweat to the surface, presenting as droplets of blood mixed with sweat. 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 may be preceded by intense headache, abdominal pain and are usually self-limiting. In some conditions, the secreted fluid is more dilute and appears to be blood-tinged, while others may have darker bright red secretions resembling blood. While the extent of blood loss generally is minimal, hematidrosis also results in the skin becoming extremely tender and fragile. Hematidrosis generally happens when a person feels intense fear or stress. Someone facing death may have this kind of fear or stress.

Case Report

A 13-year-old madrasa going girl was brought to the Department of Child and Adolescent Psychiatry of National Institute of Mental Health (NIMH), Dhaka with the complaints of recurrent bleeding from right nostril, right ear and right eye for 1.5 months. Few days before the initial episode, she experienced a hypnogogic hallucination in the form of a voice telling her days are over and she is going to die. This was followed by a few days of recurring nightmare with similar theme. Initially the bleeding was from nose, for which they consulted an ENT specialist, who examined her but could not find any definite abnormality. The bleeding did not happen for a few days, so she and her family members were relieved. Then she developed bleeding from her right eye, which happened 3 times on the same day. After 2 days gap, she developed bleeding from her right ear as well. Each of these episodes lasted for 3-5 minutes and around 5 ml of blood was lost during each of them. She developed a recurring headache at the same time since these episodes started but that did not occur exclusively during those episodes. The headache was dull aching, lasting for days, located on her forehead. She had no known history of abnormal bleeding disorders, ear infection, nasal blockade, physical trauma, fever, oral ulceration, photosensitivity, joint pain, alopecia, yellowish coloration of eyes or skin, itching, haematemesis, melaena or haemoptysis. She had no known history of taking any anticoagulant or antiplatelet drugs or food colorant. Family history of consanguineous marriage between her parents was present but no known history of bleeding disorder in her family was found. After she developed bleeding from her ear, they consulted with a medicine specialist, who referred her to a haematologist. But

after detailed history taking, physical examination and laboratory evaluation, no cause of abnormal bleeding could be detected. Then the patient was referred to National Institute of Mental Health, Dhaka as a suspected case of factitious disorder. Her laboratory investigations revealed that Hb:10.6 g/dl, total count of WBC: 5000/cmm and platelet count was normal. Bleeding time, clotting time, prothrombin time, activated partial thromboplastin time, International Normalized Ratio (INR), renal and liver function test, alpha-fetoprotein level, platelet function test, factor xiii level were within normal limit. Serum ANA, anti-dsDNA, serum c-ANCA and p-ANCA test results were also negative. Chest X-ray P/A view, X-ray PNS at OM view, CT scan of head, functional endoscopy of nose and paranasal sinuses, neck angiography, HRCT of temporal bone audiogram and tympanogram did not reveal any bleeding lesions at ear, nose, eye that could explain the symptoms.

Figure 1
Recurrent bleeding from right eye

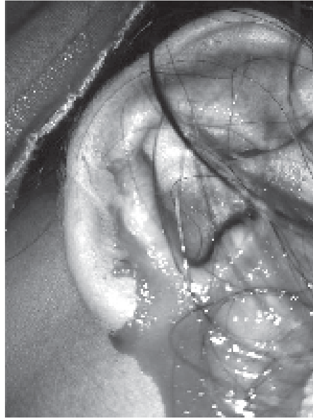

Figure 2
Recurrent bleeding from right ear

After her admission in NIMH, she continued to experience bleeding episodes, now involving left ear as well. On observation, each episode started with oozing of blood and persisted for 2-3 minutes which was painless, subsided spontaneously without leaving any bleeding point, scar or injury mark. The episodes were precipitated by a sensation of palpitation and apprehension. During bleeding episodes, blood was collected in a test tube using a cotton swab and microscopic examination of the specimen revealed the same component as normal human blood which also came positive after benzidine test. Special stains to detect hemosiderin (Prussian blue) was positive. Psychiatric evaluation revealed that she was well alert and fully conscious. Speech was low in volume and her mood was depressed, no abnormalities of thought or perception were detected. Her intelligence was within normal limit. There was no recent or past history of physical or sexual abuse. Her premorbid personality was introvert, shy and had an anxious trait. She was worried about the significance of her symptoms, thinking she might have developed some serious illness.

Depression Anxiety Stress Scale-21 (DASS-21) revealed she was severely stressed and depressed and moderately anxious. She was given propranolol (10 mg) two times daily, sertraline (50 mg) in the morning and was advised psychotherapy which included counseling, relaxation technique and cognitive behavioral therapy (CBT). At the time the case report was being written, she was still admitted and observed for improvement.

Discussion

Hematohidrosis, also known as hematidrosis, hemidrosis or hematofolliculohidrosis is an enigmatic disorder characterized by recurrent episodes of self-limiting bleeding from intact skin. It can occur at any part of the body and at several points simultaneously. Despite being extremely rare and lacking clear scientific explanation to support its existence, hematidrosis is real and has been reported for many centuries throughout the world. Diagnosis of hematidrosis can only be made if the following criteria are met: i) recurrent, spontaneous, painless and self-limited oozing of bloody discharge is witnessed and confirmed by health professionals, ii) the usual blood components are found on biochemistry 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 oozing. All of these criteria must be met in order to rule out organic bleeding disorders, self-inflicted bleeding, factitious disorder by proxy, chromhidrosis.[1,2,3] The etiology and pathogenesis of hematohidrosis remain unclear. Although experts consider extreme physical or emotional stress is the main cause of hematohidrosis, it may happen without any preceding stressful situation. In our case, intense fear was a trigger for most of the episodes.

Various causative factors, like it being component of systemic disease, vicarious menstruation, excessive exertion, psychogenic, psychogenic purpura and unknown causes have been suggested.[1] Epileptic seizure and platelet factor 3 dysfunction are also attributed to hematidrosis.[4,5] In our case, there was no evidence of self-induced injury and diagnosis was established with a demonstration of blood corpuscles in the secretion along with negative tests for bleeding diathesis. Vicarious menstruation (i.e., cyclical bleeding in extra-genital organs during a normal menstrual cycle) was ruled out in our patient since there was no relation with menstrual period. There are a number of theories regarding the explanation of hematohidrosis.

The most commonly proffered explanation relates to intensified sympathetic activation due to extreme physical or mental stress. “The fight or flight response” invoked by sympathetic activation leads to constriction of capillary vessels feeding the sweat glands. When the anxiety subsides, the blood vessels dilate to the point of rupture, leading to the passage of blood through the ducts of the nearby sweat glands and presenting as droplets of blood mixed with sweat on the intact skin surface or mucosa in almost any part of the body. Such manifestations may occur at several points simultaneously.[2,6,7] Dermal vasculitis is also concluded as a pathological basis for hematohidrosis. Stromal weakness due to defects in the dermis is another theory to explain the occurrence of hematohidrosis. According to this theory, the communication between these defects and vascular spaces in the dermis may lead to the establishment of dilated

blood centers. Whenever the positive pressure inside vascular spaces exceeds a certain level, blood will exude via follicular canals or directly into the skin surface. Subsequently, they will collapse and leave no scar. This phenomenon, which acts like a balloon, will wax and wane; thus, explaining why the bleeding are intermittent and self-limiting. The bleeding is intermittent because the vascular spaces will disappear after exuding their content but then reoccurred after the blood flow is reestablished. An immediate biopsy is important for definite diagnosis.

Biopsy during symptom free period does not reveal any blood-filled vascular spaces, intradermal bleeding or abnormality in hair follicles and sebaceous or sweat glands. The term "hematofolliculohidrosis" is proposed as it appears along with sweat like fluid and the blood pushes via the follicular canals.[2,8] Currently there is no convincing specific therapy available for this rare condition though there are reports of good response to various drugs such as anxiolytics, especially in cases triggered by extreme stress. There are some reports of successful use of propranolol.[9] Atropine sulfate transdermal patches have also been used successfully.[3] In addition, in a case with simultaneous epileptic seizure and hematidrosis, the symptoms of both were successfully resolved following the administration of anti-epileptic drug, oxcarbamazepine.[5] Hematohidrosis rarely cause serious side effects, though some people experience dehydration and anxiety. Doctors may give additional medication to treat these symptoms. Psychological counseling can also help if a person with hematidrosis has depression and anxiety. In case of our patient, beta-blocker, sertraline, supportive psychotherapy, relaxation therapy diminished the frequency of episodes.

Conclusions

In conclusion, though rare, hematohidrosis should be considered as a miscellaneous differential diagnosis of these types of bleeding episodes in a patient with normal physical & laboratory investigations. Patient should not be accused of factitious disorder which can have a tragic consequence for a family that is seeking help for their child who suffer from this very rare disorder. Further studies are needed to find out the etiology and risk factors of such condition for proper clinical management.

Anika Ferdous, Phase B MD Resident, National Institute of Mental Health & Hospital (NIMH), Dhaka, Bangladesh; **Faijul Islam,** Phase B MD Resident, NIMH, Dhaka, Bangladesh; **Taiyeb Ibna Zahangir,** Phase B MD Resident, NIMH, Dhaka, Bangladesh.

Correspondence: Anika Ferdous, Phase B MD Resident, National Institute of Mental Health & Hospital (NIMH), Dhaka, Bangladesh.
E-mail: afnbmc@gmail.com

How to cite this article: Ferdous A, Islam F, Zahangir TI. Hematohidrosis: a mysterious and rare disorder. *Arch NIMH* 2020; 3(2): 40-42.

Received 23 Aug 2020, revised 27 Oct 2020, accepted 5 Nov 2020.

References

1. Holoubek JE, Holoubek AB. Blood, sweat and fear: A classification of hematidrosis. J Med 1996; 27: 115-33.
2. Manonukul J, Wisuthsarewong W, Chantorn R, Vongirad A, Omeapinyan P. Hematidrosis: A pathologic process or stigmata. A case report with comprehensive histopathologic and immunoperoxidase studies. Am J Dermatopathol 2008; 30: 135-9.
3. Biswas S, Surana T, De A, Nag F. A curious case of sweating blood. Indian J Dermatol 2013; 58: 478-80.
4. Shen H, Wang Z, Wu T, Wang J, Ren C, Chen H, et al. Haematidrosis associated with epilepsy in a girl successfully treated with oxcarbazepine: Case report. J Int Med Res 2015; 43: 263-9.
5. Mishra KL. Bloody tears and hematohidrosis in a patient of PF3 dysfunction: A case report. Cases J 2009; 2: 9029.
6. Jerajani HR, Jaju B, Phiske MM, Lade N. Hematohidrosis-A rare clinical phenomenon. Indian J Dermatol 2009; 54: 290-2.
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. Praveen BK, Vincent J. Hematidrosis and hemolacria: A case report. Indian J Pediatr 2012; 79: 109-11.
9. Wang Z, Yu Z, Su J, Cao L, Zhao X, Bai X, et al. A case of hematidrosis successfully treated with propranolol. Am J Clin Dermatol 2010; 11: 440-3.