

## Hematidrosis: a rare clinical entity

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### Summary

Hematidrosis or hematohidrosis or hemidrosis is a very rare condition in which a human being sweats blood. Physical and psychological stresses are found to be the most frequent causes among other causes such as systemic diseases and vicarious menstruation. This is a case report of 16-year-old girl with oozing of blood from intact skin of forehead, scalp, ears, eyes and nose. Physical exertion, long journey, intense fear secondary to psychosocial stressors and academic examination were identified as precipitants in this case she was provisionally diagnosed as mixed anxiety and depressive disorder. Pharmacotherapy and psychotherapy were followed by complete remission; it is inferred from the experience that hematidrosis is a treatable condition if the underlying cause is correctly identified.

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### Introduction

Hematidrosis is a rare clinical condition of sweating of blood. It may occur when a person is suffering from extreme stress. For example, facing his or her own death.<sup>1</sup> Very few cases of hematidrosis have been reported in literature. Hematidrosis is also known as hematohidrosis and hemidrosis. The exact etiology of hematidrosis is unknown. It is believed to be a systemic disease, e.g. it has been associated with vicarious menstruation, a condition in which bleeding occurs from a surface other than the mucous membrane of the uterine cavity at a time when normal menstruation should take place. Hematidrosis has been reported with primary thrombocytopenic purpura.<sup>2</sup> In another study, a case of hematidrosis, accompanied with otorrhoea and otoerythro-sis has been reported.<sup>3</sup> Hematidrosis can also occur in the settings of excessive exertion, psychogenic and other unknown factors.<sup>4</sup> Few theories have been proposed regarding the etiopathogenesis of hematidrosis. One such school of thought says that there are multiple blood vessels around the sweat glands arranged in a net like form. It is believed that under the pressure of great stress the vessels contract. Subsequently as the anxiety passes the blood vessels dilate to the point of rupture. The blood at this point goes into the sweat glands which push the blood to the surface and manifests as droplets of blood mixed with sweat.<sup>5</sup> The term "hematofolliculohidrosis" was proposed because it appeared along with sweat like fluid and the blood exuded via the follicular canals.<sup>6</sup>

## Case Report

A 16 years college girl was brought to Child, Adolescent and Family Psychiatry Department of National Institute of Mental Health, Dhaka with complaints of recurrent episodes of bleeding for last 1 year from nose (Figure-1), ears, eyes, forehead (Figure-1), scalp, face (Figure-2), behind ears, finger pulp (Figure-3), tongue (Figure-4), sole of feet, and under the breast creases. Initially the bleeding started suddenly from nose and ear during an episode of heavy exercise in a parade ground. Then she visited ENT Department of Rajshahi Medical College Hospital and detailed ENT evaluation was done, but no abnormality was found and episode of bleeding was subsided spontaneously 6 hours later. Since then she developed 2 to 3 bleeding episodes at every 2 to 4 weeks interval from previously mentioned areas. Each episode persisted for 4 to 10 minutes which initially oozed with sweat, later on became like frank bleeding and subsided spontaneously without leaving any injury, scar mark or bleeding point.



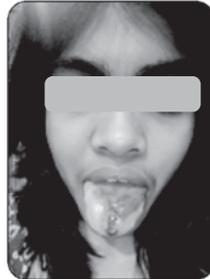
*Fig.-1: Bleeding from forehead and nose*



*Fig.-2: Bleeding from face*



*Fig.-3: Bleeding from right index finger*



*Fig.-4: Bleeding from tongue*

(Photographs are published with permission of the patient)

In each episode approximate blood amount was about 2 to 3 ml from each site. She also stated that each episode of bleeding was preceded by dull headache and abdominal pain. Episodes of bleeding were more intense and more frequent during period of intense stress especially during physical exercise, emotional upset during personal and familial conflict, during academic examination and after long journey. She had no known history of abnormal bleeding disorders, physical trauma, fever, oral ulceration, photosensitivity, joint pain, alopecia, yellowish coloration of eyes or skin, itching, hematemesis, melaena or hemoptysis. She had no known history of taking any anticoagulant or antiplatelet drug or taking food colorant. Family history of any significant bleeding disorder including consanguineous marriage among her parents was excluded. Then she was referred to Dhaka Medical College Hospital for detailed medical, hematological, ENT and dermatological evaluation. After detailed medical, hematological, ENT and dermatological clinical as well as laboratory evaluation no cause of abnormal bleeding was found. Then the patient was referred to National Institute of Mental Health, Dhaka for psychiatric evaluation as a suspected case of factitious disorder.

Her relevant investigations were done at Dhaka Medical College Hospital and results were as follows: Complete blood count- Hb% 10.5 gm/dL, platelet count normal, peripheral blood film- microcytic hypochromia, bleeding time, clotting time, prothrombin time, activated partial thromboplastin time, international normalized ratio (INR), alpha feto protein level, platelet function test, factor xiii level, eglobulin lysine test, renal and liver function test results were within normal limit. Serum ANA, anti ds DNA, serum c-ANCA & p-ANCA test results were negative. Chest X-Ray P-A view, X-Ray PNS at OM view, CT Scan of head, orbit, nose and paranasal sinuses, endoscopy of upper GIT, functional endoscopy of nose and paranasal sinuses, neck angiography, HRCT of temporal bone, audiogram and tympanogram including direct laryngoscopy did not reveal any bleeding lesion at ear, nose, throat, eye and upper GIT that could explain the previously mentioned symptoms.

After her admission in NIMH, she developed further bleeding episode for 5 times. On observation each episode started with oozing of blood from face, forehead behind ears and trunk and persisted for 2 to 3 minutes which subsided spontaneously without leaving any bleeding point, scar or injury mark from bleeding areas and each bleeding episode was painless. During bleeding episodes blood was collected using cotton swab and microscopic examination of the specimen revealed the same component as normal human blood which also came positive after benzidine test. Biopsy done during remission revealed an unremarkable epidermis, capillary sized vessels with RBC in their lumen in the dermis along with papillary dermal edema and dermal melanophages. Special stains to detect hemosiderin (prussian blue) was positive. During psychosocial evaluation the girl was alert, well oriented, comprehended and communicate relevantly. No psychotic symptom was elicited. She was euthymic and reactive. Her intelligence was within normal limit. When enquired about her school and scholastic performance her affect changed immediately. She became anxious and expressed fear about examination as well as academic performance. She had some sort of perfectionist trait since childhood. She had no recent or past history of physical or sexual abuse.

Analysis of stress with response to stress Questionnaire- Child/Adolescent version showed that child was definitely stressed. The girl fulfilled the criteria of mixed anxiety and depressive disorder apart from his bleeding problem. She had given Tab. Propranolol (20 mg) three times daily, Tab. Sertraline (100mg) daily at morning, Tab. Clonazepam (0.5 mg) daily at night and advised psychotherapy such as counseling, relaxation technique and cognitive behavioral therapy (CBT) to reduce the stress. There was complete subsidence of bleeding after 7 days and then she was discharged with advice for regular follow-up and psychotherapy at every 15 days interval in our outpatient department.

## Discussion

Hematidrosis is a condition in which capillary blood vessels that feed the sweat glands rupture, causing them to exude blood and occurs under conditions of extreme physical and emotional stress.<sup>5</sup> One author proposed the term "hematofolliculohidrosis" because it appears along with sweat fluid and blood exude via the follicular canals.<sup>6</sup> Various causative factors, like it being component of systemic disease, vicarious menstruation, excessive exertion, psychogenic, psychogenic purpura and unknown causes have been suggested.<sup>1</sup> Acute fear and in time mental contemplation are the most frequent causes, as reported in six cases in men condemned to execution, a case occurring during the London Blitz, a case involving fear of being raped, a case of fear of a storm while sailing etc.<sup>6</sup> In another Indian case report, the probable cause for hematidrosis was chronic stress, as the other causes were ruled out by detailed investigations.<sup>7</sup> Hysterical mechanism and psychosomatic disorders are also believed to induce bleeding.<sup>6</sup> psychogenic purpura is supposed to be caused by hypersensitivity to the patient's own blood on autoerythrocytic sensitization and is characterized by repeated crops of ecchymosis, gastrointestinal bleeding and hematuria.

Another type of bleeding through skin is psychogenic stigmata; a term used to signify areas of scars, open wounds or bleeding, through the unbroken skin, Patients belonging to this group were found to be frequently neurotic. The clinical findings of this type are a slight elevation of skin before prolonged oozing of blood; a pea sized bluish discoloration on patient's palms and erysipelas like lesions. In another study, a patient developed bleeding from her old scar whenever she had severe anxiety.<sup>6</sup>

The extravasated blood has identical cell components as of peripheral blood. 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 duct of sweat glands. Effect on the body is weakness and mild to moderate dehydration from the severe anxiety and blood and sweat loss.<sup>8</sup>

It has recently been proposed that there may be some defects in the dermis causing stromal weakness. These defects will communicate with vascular spaces in the dermis and they will eventually dilate and enlarge as blood filled spaces when the blood comes in. After that, they will exude the blood out by either via follicular canals or directly onto the skin surface and thus will occur whenever the positive pressures inside is enough. Later they will collapse leaving no scar. This phenomenon acts like a balloon waxes and wanes and thus explains why these bleeding episodes are sometimes intermittent and self-limiting. Immediate biopsy is important because a biopsy done after these spaces collapsed will not help in identifying them.<sup>6</sup> One study revealed some intradermal bleeding and emphraxised (obstructed) capillaries. No abnormality was found in sweat gland, hair follicle and sebaceous gland. They concluded that pathological basis for hematidrosis might be a distinctive vasculitis.<sup>8</sup> In this child no underlying systemic disease was found. Intense physical and psychosocial stressor such as physical exertion, long journey, examination fear, parental conflict all are responsible for spontaneous bleedings. Recursion was achieved with pharmacotherapy, supportive psychotherapy and relaxation technique.

## Conclusion

Both physical and mental stresses are important contributory factors which manifest in different forms, both physically and psychologically. In this girl apart from causing mental distress immediately, it also acts as an important epigenetic factor for hematidrosis. Successful treatment of this condition with beta blocker,<sup>8</sup> anxiolytic,<sup>8</sup> and antidepressant<sup>8</sup> are mentioned in the literature along with psychotherapy.

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