Hematohidrosis: A rare clinical entity

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ABSTRACT

Hematohidrosis is a rare clinical condition of sweating blood. A 13-year-old boy was presented to the department of dermatology with a history of spontaneous bleeding from skin since January 2007. During examination, it disappeared as soon as it was mopped leaving behind no sign of trauma only to reappear within a few seconds. This confirms that it was sweating of blood and not bleeding. Bleeding time, clotting time and prothrombin time was normal. Patient was diagnosed with hematohidrosis clinically by exclusion, confirmed by benzidine test, biochemical and microscopic examination of fluid. At present, no treatment is available for this condition. Etiology is unknown till date. Stress may be a precipitating factor.

Key words: Hematohidrosis, blood, sweat

INTRODUCTION

Hematohidrosis is a rare clinical condition of sweating blood. It may occur when a person is suffering from extreme stress, for example, facing his or her own death.[1] Very few cases of hematohidrosis have been reported in literature. Hematohidrosis is also known as hematidrosis and hemidrosis. The exact etiology of hematohidrosis 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. Hematohidrosis has been reported with primary thrombocytopenic purpura.[2] In another study, a case of hematidrohosis, accompanied with otorrhea and otoerythrosis, has been reported.[3] Hematohidrosis can also occur in the settings of excessive exertion, psychogenic and other unknown factors. It has also been reported in an 8-year-old child just as in this case without any specific pathology.[4]

Few theories have been proposed regarding the etiopathogenesis of hematohidrosis. 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.^[5] The term "hematofolliculohidrosis" was proposed because it appeared along with sweat-like fluid and the blood exuded via the follicular canals.^[6]

CASE REPORT

Hereby, we report a case of 13-year-old boy with history of spontaneous bleeding from skin and mucous membranes since January 2007. He bled daily, more so during school hours of 9-11 am and this lasted for around 1 minute. It recurred after 5 minutes and the patient bled continuously or intermittently followed by spontaneous cessation.

There was no past history or family history of bleeding tendencies, nor of any systemic diseases. There was no evidence of any trauma, purpura, scratch marks or any sign suggestive of bleeding.

On examination, there was oozing of blood stained fluid from face [Figure 1], arm [Figure 2] and trunk. On mopping, it disappeared immediately leaving behind no signs of trauma, nor any disease only to appear again within few seconds. This fact confirmed it as sweating of blood as opposed to bleeding. Patient was referred to ENT department for history of bleeding from external ear.



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Figure 1: Sweating blood from face



Figure 2: Sweating blood from arms

No sign of bleeding, injury or any systemic disease was found on examination. He was also referred to psychiatric department to rule out any underlying stress. He was investigated for complete blood count, urine routine, liver function tests and renal function tests which were all normal. Tests for bleeding tendencies were within normal range. Bleeding time was 2.15 min [normal range (N)- 2–7 min], clotting time was 6 min (N-5–10 min), prothrombin time 15.1 (control- 15.6 sec), activated

partial thromboplastin- 31.8 sec (control- 32.0 sec), platelet count was 3.3 lakhs (N- 1.5–4.5 lakhs/cu.mm) and α -feto protein levels were 1.0 IU/ml (N- 0–8 IU/ml). Platelet function tests, factor XIII and Eglobulin Lysin test time were also normal. Biopsy of skin was not taken as patient was very anxious during attack. Child apperception test suggested underlying emotional conflicts.

Computed tomography (CT), intracranial and neck angiography and magnetic resonance imaging of brain were normal. HRCT of temporal bone, audiogram and tympanogram were normal.

Diagnosis of hematohidrosis was made clinically, and confirmed by benzidine test in which hemoglobin in blood reacts with hydrogen peroxide liberating oxygen, which then reacts with organic reagent producing a green to blue-colored compound.

Microscopic examination of fluid showed plenty of RBCs indicating blood in sweat. Biochemical examination of fluid showed component of blood and sweat. K⁺ level was high because of lysed RBC.

Hemochromogen test was not done because of non-availability.

Patient was advised rest, meditation, change of lifestyle and other relaxation techniques.

Patient felt better with fewer recurrences when he was followed up after 15 days.

He was given Alprazolam (0.25 mg) once daily for 10 days, which was then stopped. We unsuccessfully tried 20% aluminium hexahydrate in alcohol to control sweating during school hours. The patient then decided to opt for alternative medicinal system along with breathing exercises and reported no recurrence until he was on that treatment. However, it recurred as soon as he stopped that treatment.

DISCUSSION

Hematohidrosis also known as hematidrosis, hemidrosis and 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 or emotional stress. [5] One author proposed the term "hematofolliculohidrosis" because it appeared along with sweat-like fluid and blood exudes via the follicular canals. [6]

Various causative factors, like it being component of systemic disease, vicarious menstruation, excessive exertion, psychogenic, psychogenic purpura and unknown causes have been suggested. [1] Acute fear and intense 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.^[6] In another Indian case report, the probable cause for hematohidrosis was chronic stress, as the other causes were ruled out by detailed investigations.^[7] Hysterical mechanisms and psychosomatic disorders are also believed to induce bleeding.^[6] Psychogenic purpura is supposed to be caused by hypersensitivity to the patients' own blood or autoerythrocyte sensitization and is characterized by repeated crops of ecchymoses, gastrointestinal bleedings 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 palm and erysipelas-like lesions. In another study, a patient developed bleeding from her old scars whenever she had severe anxiety. [6]

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 ducts of the sweat glands. Effect on the body is weakness and mild to moderate dehydration from the severe anxiety and both blood and sweat loss.^[8]

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 on to the skin surface and this will occur whenever the positive pressure 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 collapse will not help in identifying them. [6] One study revealed some intradermal bleeding and emphraxised (obstructed) capillaries. No abnormality was found in sweat glands, hair follicles and sebaceous glands. They concluded that pathological basis for hematohidrosis might be a distinctive vasculitis. [8]

We are unable to comment on the efficacy of the alternate medicinal systems. as the patient was on multiple therapies. Spontaneous remission cannot be ruled out. We report this case for its rarity. At present, no specific treatment is available for this condition.

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