A case report of hematidrosis: blood, sweat, and fear

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ABSTRACT

Background: Hematidrosis is an extremely rare condition in which the patient spontaneously starts sweating blood. Even though, this disease has existed for many centuries, due to its rarity and perceived religious connection made little progress in correctly evaluating its pathogenesis. Therefore, rather than getting the medical and emotional help these cases need, such cases often end up at the doorsteps of shamans, especially in developing countries.

Case Presentation: Here, we describe a case of hematidrosis and its clinical presentation and treatment outcome. A 10-year-old female patient presented with complaints of episodic bleeding from the skin, predominantly from the trunk, oral cavity, and forehead. Blood investigations, including coagulation profile, platelet antigen study, etc. were completely normal. The patient was treated with propranolol and atropine transdermal patches, which showed improvement in her condition, and the bleeding eventually stopped.

Conclusion: Standardized diagnostic and management criteria is needed to be developed for treating such patients better. Reporting such cases of hematidrosis and discussing them will eventually make disease management more uniform and bring more patient awareness to this frightening disease.

Keywords: Hematidrosis, hematohidrosis, blood in sweat.

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Background

Etymology: *Greek: Haïma*, meaning Blood; *Hïdrös*, meaning Sweat.

Hematidrosis is an extremely rare condition in which the patient spontaneously starts sweating blood. Literature and research regarding such cases are not easily available due to their rarity. It often occurs in patients suffering from stress and anxiety. Here, in this case report, we try to understand the clinical presentation, associated fear and stigma, investigations, and treatment options for hematidrosis.

There have been few historical references made to this disease, However, most such references were connected to religious explanations, rather than scientific ones. In the Bible, it is said that Jesus Christ experienced hematidrosis before the crucifixion, as Luke 22:44 says, "*…and being in anguish, he prayed more earnestly, and his sweat was like drops of blood falling to the ground.*"

One of the earliest mentions of this disease has been done by Leonardo da Vinci in the 15th century when he described a soldier who sweats blood in stressful situations.

Even in today's modern era, sweating blood without injury often raises fear and distress in patients and their families.

Duan et al. [1] mentioned such a case associated with primary thrombocytopenic purpura and Migliorini has stated a case of Hematidrosis otorrhea with otoerythrosis [2]. Herein, we present a case report of isolated hematidrosis without any other associated systemic manifestations.

Case Presentation

A 10-year-old female presented with complaints of episodes of bleeding from the skin for the last 1 year. The primary areas involved were the trunk - near the umbilicus, forehead, oral cavity, and groin (Fig. 1 and 2). These episodes were intermittent with a frequency of one episode every 1-2 days and occasional multiple episodes in a day as well.

The bleeding was peculiar in three ways:

1. Bleeding was from intact skin or mucosa.
2. Bleeding was episodic, spontaneous, and self-limiting, lasting only a few minutes.
3. After the bleeding stops, there will not be any visible cuts or abrasions or trauma to the skin.

These episodes of bleeding were not associated with or preceded by fear, stress, or other precipitating factors, and were spontaneous. At the beginning of each episode,

profuse sweating will start, which will soon be followed by bright red-colored blood coming out of the skin. In most episodes, only a single site was involved, with no multiple areas of bleeding in the same episode. After a few minutes, the bleeding used to stop eventually on its own, with no subsequent symptoms. Each episode will last for 7-8 minutes, and the bleeding was self-limiting. The general condition of the patient remained all right with normal vitals.

There was no history of usage of anti-coagulants or other blood thinners. There was no major medical or surgical history. Family history was as well, non-significant.

Upon the general and systemic examination, no major abnormalities were detected. The area of bleed appeared normal post-bleeding and had no tenderness or visible swelling (Fig. 3). Upon further examination, bleeding would not occur if the concerned area were squeezed or compressed.

The examination of secretion revealed that it was bright red in color, with no clots or clumps in it, and its viscosity was slightly less than the blood. Microscopic examination showed multiple red blood cells with few white blood cells as well. All her blood investigations were normal (Table. 1), with a normal coagulation profile, normal Liver Function Tests (LFTs), normal Hb electrophoresis (Table. 2), normal platelet counts, and Table. 3, 4 and 5, and Peripheral Blood Smear (PBS). (Table. 3, 4 & 5). The patient had a normal psychological evaluation with no signs and symptoms of mental disorders, mental retardation, anxiety, or other psychological illnesses.

The patient was at first treated with tab propranolol 10 mg once a day, which showed partial improvement, as bleeding episode frequency was decreased from once a day to one episode in 3-5 days. The patient was then given atropine sulfate transdermal patches which further improved the symptoms and the bleeding episodes stopped completely.

Discussion

Hematidrosis is also sometimes referred to as hematohidrosis, hemidrosis, or hematofolliculohidrosis. Since hematidrosis cases are quite rare, the available literature is very scarce. Shafique et al. [3] described a few cases of hematidrosis, in which the majority of cases (83%) were of female gender and of age <18 years. Some patients had shown prodromal symptoms like headache, nausea, and local tingling sensation before the bleeding episode. Case series published by Tshifularo [4] and Manonukul et al. [5] noted that psychological stress was an associated condition and was probably a causative factor.

It is extremely crucial that before making the diagnosis of hematidrosis, we must differentiate it from either self-inflicted/otherwise accidental skin injuries or other pathologies with similar presentation such as vasculitis, chromhidrosis

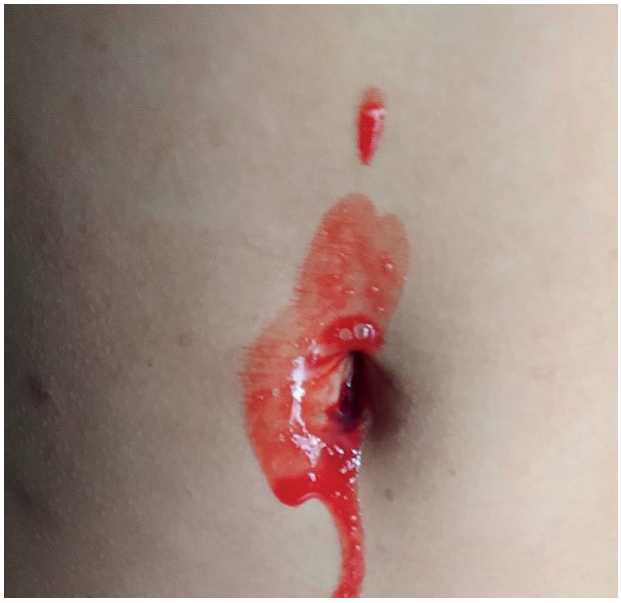

Figure 1. *Bleeding around umbilicus.*

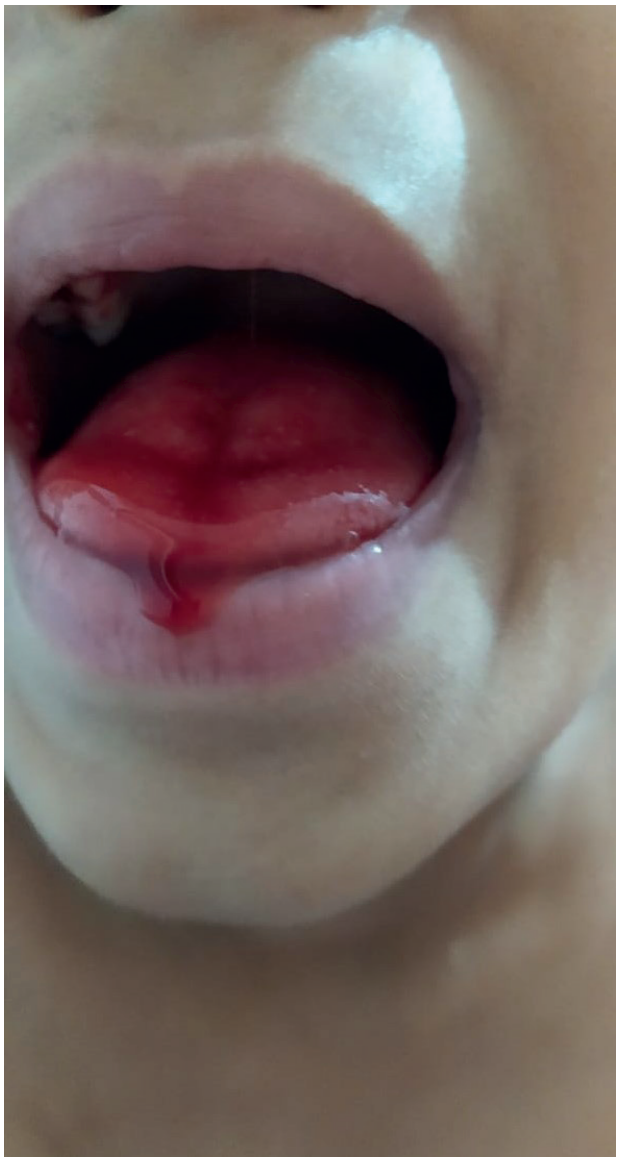

Figure 2. *Bleeding from oral cavity.*

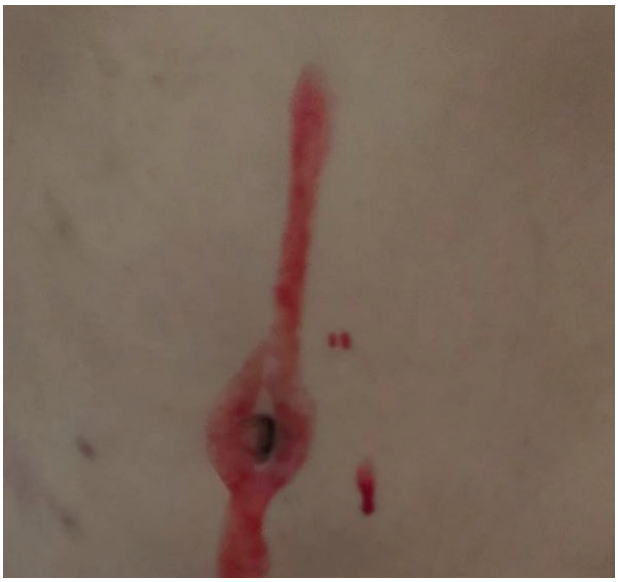

Figure 3. *Bleeding.*

Table 1. *CBC report of the patient.*

PARAMETERS	PATIENT'S VALUES	REFERENCE RANGE
Hemoglobin (g/dl)	13.0	11.5-15.5
RBC count (× 1012/l)	4.77	4.0-5.2
Hematocrit (%)	38.0	35-45
MCV (fl)	79.7	77-95
MCH (Pg)	27.3	25-33
MCHC (g/dl)	34.2	31-37
RDW (%)	13.2	11.5-14.5
WBC Count (per cmm)	7,340	5,000-13,000
Neutrophils (%)	39.9	30-70
Lymphocytes (%)	48.2	20-50
Monocytes (%)	6.4	0-10
Eosinophils (%)	5.0	0-4
Basophils (%)	0.5	0-1
Immature granulocytes	0.1	0-0.5
Absolute neutrophil count	2,930	1,800-7,000
Absolute lymphocyte count	3,530	1,200-5,000
Absolute monocyte count	470	100-400
Absolute eosinophil count	370	20-300
Absolute basophil count	40	0-100
Neutrophil lymphocyte ratio	0.83	0.1-4.5
Platelet count	301,000	170,000-450,000
MPV	8.1	7.2-11.7

Table 2. *Hb electrophoresis of the patient.*

PARAMETERS	PATIENT'S VALUES	REFERENCE RANGE
HbF level	0.8%	0-4
HbA2	3.0	0-3.5
E - Window	0.0	0-15
D - Window	0.0	0-15
Sickle cell - Window	0.0	0-15
C - Window	0.0	0-15

Normal Hb electrophoretic pattern. No evidence of hemoglobinopathies.

Table 3. *Clot retraction test of the patient.*

PARAMETERS	PATIENT'S VALUES	REFERENCE RANGE
Clot retraction test	Good	-
C - Window	0.0	0-15

Table 4. *Coagulation profile of the patient.*

PARAMETERS	PATIENT'S VALUES	REFERENCE RANGE
Activated partial thromboplastin time	34.8 seconds	25.4-38.4 seconds
Mean - APTT (Seconds)	31.3	25.4-38.4
Prothrombin time (Seconds)	11.7	10.5-13.5
MNPT (Seconds)	11.6	10.5-13.5
INR	1.01	
Ristocetin-induced platelet aggregation	Normal platelet aggregation	Normal platelet aggregation

Normal coagulation profile.

Table 5. *Platelet antigen study (Gb IIb/IIa and Gb Ib) of the patient.*

PARAMETERS	PATIENT'S VALUES
CD41 Antibody	100% Bright intensity
CD61 Antibody	100% Bright intensity
CD42b Antibody	100% Bright intensity

No phenotypic evidence of deficiency of platelet membrane Gp IIb/IIa (Glanzmann's thrombasthenia) or Gp Ib (Bernard Soulier Syndrome).

(colored sweat), or other connective tissue disorders where vascular pathology leads to bleeding from the skin.

The exact etiopathogenesis of hematidrosis is unknown, but few hypotheses have been proposed. One hypothesis presented by Uber et al. [6] says that sympathetic stimulation causes constriction, thus raising vascular pressure and then dilatation of capillaries surrounding the sweat glands, causing blood to enter sweat ducts and then appear along with sweat. The fact that beta-blocker has been a treatment of choice in our and many other hematidrosis cases, makes this hypothesis more plausible. Another hypothesis, presented by Manonukul et al. [5] says that defects in the dermis cause its direct communication with the vascular spaces filled with blood, thus allowing blood to pass into follicular canals and eventually over the skin surface. Zhang et al. [7] suggested that obstructed peripheral capillaries along with vasculitis are the main pathology in hematidrosis. However, many cases reported by other authors show normal skin

biopsies and no features of vasculitis, thus vasculitis as a causative factor for hematidrosis appears less convincing.

Management options range from beta-blockers, atropine patches, anxiolytics, Selective Serotonin Reuptake Inhibitors (SSRIs), tricyclic antidepressants, psychotherapy, etc. Some cases have shown remission without any intervention, while some have not responded at all with treatment options.

A thorough evaluation of the etiopathology of this disease is essential in developing a comprehensive management algorithm for the disease.

Conclusion

Hematidrosis can be a frightening and fearful experience for patients and their families, which leads to a repeated unnecessary and futile exercise of excessive hematological evaluation which mostly comes normal, often making patients, and in some cases, clinicians too, more stressed, unable to find the explanation. Although the condition is benign and has no major local or systemic consequences, the sight of blood in sweat usually makes the patient petrified. The diagnosis of hematidrosis should be done as a diagnosis of exclusion, only after ruling out other possible disease pathologies. Reporting such cases of hematidrosis and discussing them will eventually make disease management more uniform and bring more patient awareness to this frightening disease.

What is new?

Hematidrosis is an extremely rare condition in which the patient spontaneously starts sweating blood. Its pathogenesis, diagnosis, and treatment options are not been very well studied and well discussed so far. Therefore, rather than getting the medical and emotional help these cases need, such cases often end up at the doorsteps of shamans, especially in developing countries.

List of Abbreviations

CBC	Complete Blood Counts
Hb	Hemoglobin
HbF	Hemoglobin (Fetal)
INR	International Normalised Ratio
LFTs	Liver Function Tests
MCH	Mean Corpuscular Hemoglobin
MCHC	Mean Corpuscular Hemoglobin Concentration
MCV	Mean Corpuscular Volume
MNPT	Mean Normal Prothrombin Time
MPV	Mean Platelet Volume
PBS	Peripheral Blood Smear
RBC	Red Blood Cells
RDW	Red-cell Distribution Width
SSRI	Selective Serotonin Reuptake Inhibitors
WBC	White Blood Cells

Conflict of interest

The authors declare that there is no conflict of interest regarding the publication of this case report.

Funding

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Consent for publication

Written consent was obtained from the parent of the patient.

Ethical approval

Ethical approval is not required at our institution for publishing an anonymous case report.

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Summary of the case

1	Patient (gender, age)	Female, 10-year-old
2	Final diagnosis	Hematidrosis
3	Symptoms	Blood in sweat
4	Medications	Propranolol, Atropine sulfate
5	Clinical procedure	Propranolol 10 mg once daily, along with atropine sulfate transdermal patch
6	Specialty	Dermatology, Internal medicine