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RESEARCH ARTICLE

A13-YEAR-OLD FEMALE WITH HEMOLACRIA AND HEMOPTYSIS

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ABSTRACT

Hematidrosis is an exceptionally rare disease that was associated with religious beliefs, high blood pressure, menses, stress and systemic disease. This disease has been described as a stigmata, which is a term used by Christians to explain body marks, sores, pain, and bleeding in locations consistent with the crucifixion wounds of Jesus Christ. These wounds are typically on the hands, wrists, and feet. However, pathophysiology and etiology are still largely unknown. A complete patient's medical and family history was obtained, physical examination and a detailed laboratory were conducted. A spectrum of autoimmune and oncological diseases were assayed. Ultrasounds, radiography, echocardiography, endoscopy, esophago-gastro-duodenoscopy were performed for final diagnosis. The patient's past medical history were unremarkable. Physical examination was unexceptional for any abnormal findings. All of the labs were reported to be within normal limits. All the assays for autoimmune diseases were negative. The investigation at multiple facilities by pediatric nephrologists, hematologist-oncologists, immunologists, psychiatrists, and endocrinologists did not establish diagnosis. Bronchoscopy and gastroduodenal endoscopy were normal. The patient was discharged without diagnosis. After repeated bleedings a hematologist diagnosed this patient with hematidrosis. The diagnosis is consistent with this patient as she seemed to exhibit issues when she was stressed and her blood pressure increased.

INTRODUCTION

Hematidrosis, a bleeding disorder also called blood sweat, is an extremely rare condition in which a human sweats blood (1,2). Blood also oozes from the forehead, umbilicus, and other skin surfaces causing nosebleeds, blood stained tears, otoerythrosis and vicarious menstruation (3). There are many factors believed to contribute to this disease. One publication indicates that stress and strenuous exercise may proliferate the issue, and it may occur in individuals with underlying bleeding disor (4). Although the exact pathologic mechanism remains unclear, it has been suggested that dermal defects may lead to blood-filled spaces that would exude into follicular canals or directly to the skin surface (5). Biopsies immediately proceeding the bleeding are remarkable for blood-filled cavities surrounding the ducts that open directly into the follicular canals or directly onto the skin. Several studies have indicated that patients' symptoms dramatically resolve when treated with beta blockers. It has been suggested that sympathetic nerve activity may play a role in these patients' bleeding events. As such the beta adrenergic receptor antagonists are effective for treating these patients (6). The differential diagnosis list included factitious disorder, and bleeding diathesis. Here we report a case of a 13-year-old girl who frequently bled from the scalp, eyes and ears.

MATERIAL AND METHODS

A complete medical and family history was obtained. A comprehensive physical examination was performed and a detailed laboratory analysis of peripheral blood and urine were conducted. The antibodies for the spectrum of autoimmune and oncological diseases were assayed. Non-invasive methods (ultrasounds, radiography, and echocardiography) as well as invasive methods (endoscopy, esophago-gastro-duodenoscopy), were used as diagnostic methods to obtain the final diagnosis.

RESULTS

The patient was examined in five different hospitals for the bleeding from the eyes, ears and nose, and for coughing up blood (Figure 1).

Figure 1 Bleeding form the eyes, ears, nose and coughing up blood

The patient's past medical history was unremarkable for any illnesses or hospitalizations. Physical examination was unexceptional for any abnormal findings. All of the patient's laboratory results were reported to be within normal limits, Complete Blood Cell Count (Table 1and Table 2), and Bleeding times (Table 2).

Table 1 Complete Blood Cell Count

Result	Value	Ref Range
Fibrinogen	331	204 - 475 mg/dl
WBC	9.3	4.0 - 10.5 K/µl
RBC	4.46	4.1 - 5.1 M/µl
Hemoglobin	13.2	12.0 - 16.0 g/dl
Hematocrit	37.7	36 - 46%
MCV	84.5	78 - 102 fl
MCH	29.6	27 - 34.6 pg
MCHC	35	33 - 37 g/dl
ROW	12.5	11.5 - 14.5%
Platelet Count	246	130 - 400 K/µl
MPV	9.6	9.4 - 12.4 fl
NRPC	0	0 - 0.2%
Absolute NRPC	0	%
Seg	68.2	%
Lymph	27.6	%
Monos	2.9	%
Eos	0.9	%
Basa	0.2	%

Table 2 Bleeding times

Result	Value	Ref Range
Immature Granulocyte	0.2	%
Absolute Neut	6.3	1.8 - 8.0 K/ul
Absolute Lymph	2.6	1.2 - 6.0 K/ul
Absolute Mono	0.3	0 - 0.8 K/ul
Absolute Eos	0.1	0 - 0.4 K/ul
Absolute Basophils	0	0 - 0.2 K/ul
Absolute Immature Gran.	0	0.00 - 0.02 K/ul
Pt (Patient)	13.7	11.6 - 15.2 sec
INR	1	Ratio
PTT	25.8	24.0 - 37.0 sec

The urine analysis also came back within normal limits (Table 3).

Table 3 Urine analysis

Result	Value	Ref Range
Glucose, Ur Bilirubin	Neg	Neg MG/dL Neg
Ketones	Neg	Neg MG/dL Neg
Specific Gravity (urine)	1.015	1.005-1.030
Blood, Urine	Neg	Neg
pH, Urine	7	5.0-8.0
Protein, Urine	Neg	Neg G/mgL
Urobilinogen, Ua	<2.0	>2.0 mg/dL
Nitrite, Urine	Neg	Neg
WBC Esterase/Urine	Neg	Neg

All the assays for autoimmune diseases were negative (Table 4).

Table 4 Antibody Assays

Antibody	Results	Normal Titer
Sjogren's Anti-SSA (Ro)Ab	Negative	<1
Sjogren's Anti-SSB (La) Ab	Negative	<1
Anti-Smith (Sm) Ab	Negative	<1
Anti-Sm/RNP Ab	Negative	<1
Anti-Scleroderma-70 Ab	Negative	<1
Anti-Jo-1 Antibody	Negative	<1
Double Stranded DNA Ab	Negative	<1
Anti-Centromere Ab	Negative	<1
Ribosomal Ab	Negative	<1
Histone Antibody	Negative	<1
Factor VIII Activiy	Positive	

Bronchoscopy (Figure 2A) and gastro-duodenal endoscopy (Figure 2B) were also reported as normal.

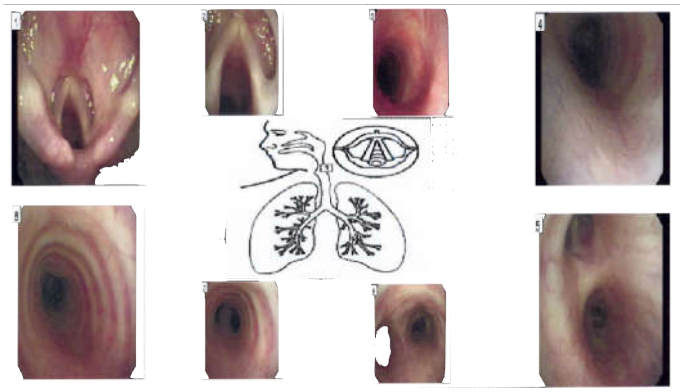

Figure 2A Bronchoscopy findings

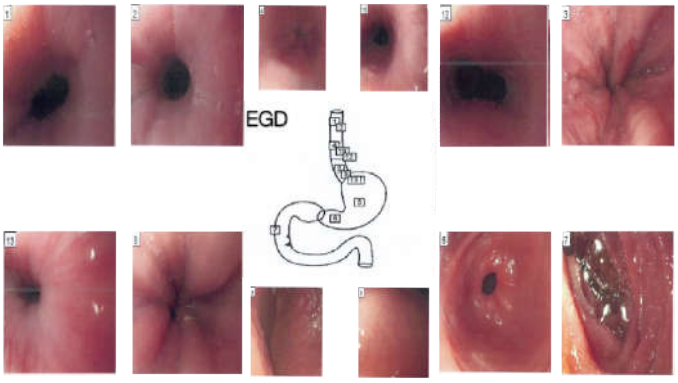

Figure 2B Esophago-gastro-duodenal endoscopy

After repeated bleedings a hematologist diagnosed this patient with hemathidrosis.

DISCUSION

Hematidrosis or Hematohidrosis (also known as hemidrosis) is a condition in which capillary blood vessels around sweat glands rupture, occurring under conditions of extreme physical or emotional stress causing them to exude blood (5). This patient was a healthy 13-year-old female with no prior medical history who played softball and basketball when she began to exhibit sudden insidious onset of headache and hemoptysis and was transported to the hospital for the first time. The patient's past medical history was unremarkable for any illnesses or hospitalizations aside from a tonsillectomy and "benign hypertension". Different causative factors have been proposed as acute fear and high blood pressure (3). Blood pressure of a patient with hematidrosis can increase incredibly and when anxiety levels go up, they sweat blood. This happens usually when an extremely dangerous condition occurs to him or her, and at the time of deepest stress. When a person is facing his death or sometimes when they get too stressed about something they may get blood as sweat. Although, historically it was described to occur under conditions of extreme physical or emotional stress some reports have shown that it may occur without stressful situations (7). Vicarious menstruation was reported to be accompanied with hemathidrosis (3). Our patient started her period at 12 and has had regular periods since this time and was not sexually active. In our case, menstruation was excluded as a causative factor because bleeding occurred between menstrual periods. She does not smoke, drink or use illicit drugs. Factitious disorder is also possible, although the prototypical red flags were not present

in her interactions with family. Additionally, bright red secretions from the ear canals and eyes were witnessed by the nurses in the ED before the patient was seen. The nurses in the ED also witnessed pink spots when she coughed into a rag when she was in intake. Factitious disorder was ruled out by in house psychiatric team. Blood biochemistry and Complete blood count was normal, with normal coagulation screening tests. Von-Willebrand factor and platelet aggregation test revealed no abnormality. Underlying autoimmune conditions were excluded because Assay of anti-nuclear antibody revealed no abnormality. Hematidrosis can be a component of systemic disease but in this patient physicians were able to exclude all autoimmune diseases, coagulopathies, bleeding diathesis, psychiatric disorders, cancers, endocrinopathies and nephropathies. Patient was temporarily placed on Propranolol to control her blood pressure because there is some indication in the literature that this disease process may be associated with hypertension (8, 9). However, long term anti-hypertenaives were deemed unnecessary, as high blood pressure was not considered to be the causative factor of hemathidrosis in this case. She was cleared to return to normal activity from a blood pressure standpoint. Bleeding diathesis is also possible, but this is less likely given that the patient did not experience gum or other mucosal bleeding and all of the bleeding and coagulation studies came back normal. Stigmata is also a plausible differential, but the patient does not have body marks, sores, or sensations of pain in locations corresponding to the crucifixion consistent with this disorder. This patient was provided a provisional diagnosis of hematidrosis. All other possibilities were exhausted. A factitious disorder is possible here although the prototypical red flags are not evident in her reactions with her family and staff. Additionally, bright red secretions from the ear canals and eyes were witnessed by the nurses in the Emergency Department before the patient was seen. The nurses also noted pink spots in a wash cloth when she coughed. Bleeding diathesis is also possible, but this is less likely given that the patient did not experience gum or other mucosal bleeding and all of the bleeding and coagulation studies came back normal. Stigmata is also a plausible differential, but the patient does not have body marks, sores, or sensations of pain in locations corresponding to the crucifixion consistent with this disorder.

CONCLUSION

This patient was provided a provisional diagnosis of hematidrosis. All other possibilities were exhausted. Physicians were able to exclude all auto immune diseases, coagulopathies, bleeding diathesis, psychiatric disorders, cancers, endocrinopathies, and nephropathies. This is an extremely rare disease process that has been described as a stigmata. However, it has also been associated with several other factors such as hypertension, menses, and stress. The pathophysiology and etiology are still largely unknown. There are a number of theories but there is not definitive data. The patient was placed on Propranolol to control her blood pressure because there is some indication in the literature that this disease process may be associated with hypertension

References

1. Tshifularo M. Blood otorrhea: blood stained sweaty ear discharges: hematohidrosis; four case series (2001-2013). *American Journal of Otolaryngology*. 2014; 35 (2):271-3.
2. Manononukul J, Wisuthsarewong W, *et al*. Hematidrosis: a pathologic process or stigmata. A case report with comprehensive histopathologic and immunoperoxidase studies. *Am J Clin Dermatol* 2008; 30 (2): 135-139
3. Holoubek JE, Holoubek AB. Blood, sweat and fear: A classification of hematidrosis. *Journal of Medicine*. 1996; 27 (3-4): 115-33
4. Saugato B, Trupti S, Abhishek D, Falguni, N. A Curious Case of Sweating Blood. *Indian J Dermatol*. 2013; 58(6): 478-480
5. Souad R, Khalid K, Mohamed B, Rachid A. Hematohidrosis-a rare clinical phenomenon. *Indian J Dermatol*. 2009; 54(3): 290-292.
6. Zhaoyue W, Ziqiang Y, *et al*. A Case of Hematidrosis Successfully Treated with Propranolol. *Am J Clin Dermatol*. 2010; 11 (6): 440-443.
7. Varalakshmi B, Doshi V, Sivalingam D, Nambi S. The story of a girl with weeping blood: Childhood depression with a rare presentation, *Indian J Psychiatry*. 2015; 57(1):88-90
8. Wang Z, Yu Z, Su J, Cao L, Zhao X, Bai X, Zhan S, Wu T, Jin L, Zhou P, Ruan C. A Case of Hematidrosis Successfully Treated with Propranolol. *Am J Clin Dermatol*. 2010; 11(6):440-443
9. Mora E, and Lucas J. Hematidrosis: Blood sweat. *Blood*. 2013; 121(9):149

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