

Hematidrosis: A Fascinating Phenomenon— Case Study and Overview of the Literature

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Semin Thromb Hemost 2018;44:293–295.

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As previously explored in this journal, the investigation of bleeding symptoms involves a systematic investigative approach that includes investigation of patient and family history, physical examination, and laboratory testing.¹ Although this is also true of an investigation into pediatric bleeding, there are special considerations required, including the reduced period of capture for personal history, and fewer hemostatic challenges.² Sometimes, such investigations prove fruitless and do not uncover a true bleeding diathesis. Hematidrosis is one such example.

Hematidrosis is a fascinating disorder characterized by blood oozing from the intact skin and mucous membranes in the absence of a bleeding problem. It frequently affects young girls (i.e., typically between 9 and 13 years) under stressful situations, and probably suffering from an underlying anxiety disorder. These patients usually undergo a thorough and expensive hematology evaluation aimed to identify a possible underlying bleeding disorder, but which fails to provide the answer. Here, we report a case of a 9-year-old girl exposed to bullying at school, which leads to recurrent bleeding episodes.

The young patient had no prior medical or psychological illness and presented to our clinic following a 5-month history of recurrent spontaneous blood loss from skin, scalp, ear, mouth, and eyes (► **Fig. 1**). Her parents claimed that the episodes happen twice weekly on average, mostly in the evening, and with no clear precipitating factors. Each episode lasted for 1 or 2 minutes and was usually self-limited. Family history was negative for any bleeding disorders. She was evaluated by her family physician and a local hematologist, and a workup for common bleeding disorders was unrevealing (► **Table 1**). The patient also underwent upper gastrointestinal endoscopy because some of these episodes were accompanied by bleeding from her mouth. The endoscopy and biopsy were consistent with celiac disease, which was later confirmed by serological testing. Nevertheless, no clinical signs of celiac disease could be recorded. Upon her evaluation in our clinic, her vital signs and growth chart were within normal limits,

and physical examination was unrevealing; specifically, there were no signs of self or secondary-inflicted injuries (scars, fresh cuts, scratches, or wounds). On further questioning, the parents reported that these episodes did not occur on holidays and usually happened on the days she attended school. The patient admitted that she always felt intimidated and emotionally abused by her school colleagues. A sampling of the fluid discharge confirmed the presence of red and white blood cells along with epithelial cells.

Hematohidrosis or hematidrosis is a rare and largely mysterious phenomenon, with only a few case reports having been published.^{3–14} Reported patients are usually young girls,^{3,5} and present with blood oozing from intact skin or mucous membranes in the setting of stress and anxiety.¹¹ The episodes last for few minutes,³ and resolve almost spontaneously.⁵ The workup for bleeding disorders is usually normal, and the examination of the bloody fluid demonstrates the presence of blood elements. It is important to carefully examine the skin to rule out self-inflicted or secondary injuries (e.g., potentially pointing to Munchausen's syndrome or Munchausen's by proxy) and to also differentiate this entity from chromhidrosis (color pigment in sweat), vasculitis, scurvy, or other connective tissue disorders (where vascular fragility can lead to easy bleeding). Some of the reported cases underwent biopsy of the oozing areas with active bleeding and did not show any histopathological findings.^{4,5} Although the frightening presentation of hematidrosis leads to immediate medical evaluation in most of the cases, the diagnosis is often delayed because the condition is a rare and frequently overlooked phenomenon. Frequently, patients will have extensive evaluations including hematology consultation before reaching a correct diagnosis. The etiology remains mostly unknown. Stress-induced vasoconstriction (through adrenergic stimulation) in the small blood vessels around the sweat glands and eventually leading to vascular rupture and blood extravasation with the sweat is the proposed cause.^{10,11} The fact that most cases

published online
December 8, 2017

Issue Theme Editorial Compilation V;
Guest Editors: Emmanuel J. Favaloro,
PhD, FFSc (RCPA), and Giuseppe
Lippi, MD.

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Publishers, Inc., 333 Seventh Avenue,
New York, NY 10001, USA.
Tel: +1(212) 584-4662.

DOI <https://doi.org/10.1055/s-0037-1608905>.
ISSN 0094-6176.



Fig. 1 Spontaneous blood loss from skin, scalp, ear, mouth, and eyes during one of the bleeding episodes.

Table 1 Summary of the relevant investigations performed as a part of the initial bleeding workup

Test/analyte	Patient result (normal range)
Platelets	$241 \times 10^9/L$ ($150\text{--}400 \times 10^9/L$)
Prothrombin time	12.9 s (12.3–14.2 s)
Activated partial thromboplastin time	31.4 s (30.5–40.4 s)
Factor VIII activity	85% (60–150%)
Factor IX activity	90% (60–150%)
von Willebrand factor: Ristocetin cofactor activity	100 IU/dL (50–200 IU/dL)
von Willebrand factor: Antigen	70 IU/dL (50–150 IU/dL)
Platelet aggregation (adenosine diphosphate, collagen, arachidonic acid, thrombin, epinephrine, ristocetin) recorded by light transmission	Normal
PFA-100 (C/Epi cartridge)	112 s (78–199 s)

Abbreviations: C/Epi, collagen/epinephrine; PFA-100, platelet function analyser-100.

respond to β adrenoreceptor blockers and/or anxiolytics makes this theory plausible.^{4,10} Our patient was treated with propranolol 10 mg twice daily and her symptoms resolved. As in all the reported cases, an underlying cause usually precipitates the episodes of anxiety that lead to bleeding. The trigger in our case was identified as bullying at school; the patient and her parents were referred to psychiatry and social services for further help. The association of hematomatosis with celiac disease in our case might or might not be significant.

To conclude, hematomatosis is an interesting bleeding phenomenon where a psychological trigger (rivalry, scolding, punishment, bullying, etc.) leads to a sequence of events culminating in bleeding from intact skin and mucous membranes. This can be a frightening experience for the patient and

family and often leads to extensive unnecessary testing and sometimes even to dangerous therapeutic intervention (e.g., a case was treated with fresh frozen plasma several times⁴). Reporting such enigmatic cases might help educate the public as well as the medical society about a relatively simple yet frightening phenomenon that can exhaust resources and even lead to unnecessary therapeutic or diagnostic interventions. In the end, we decided not to pursue additional invasive investigations above those already undertaken (→ **Table 1**) based on our evaluation of this case, successful treatment with propranolol, and our conclusion that the case was indeed that of the rare condition hematomatosis.

Conflict of Interest
None.

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