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

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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. Reduced period of capture for personal history, and fewer hemostatic challenges.2 This is also true of an investigation into pediatric bleeding, and a workup for common bleeding disorders was unrevealing (Fig. 1). 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). 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.

Hematidrosis or hematidrosis is a rare and largely mysterious phenomenon, with only a few case reports having been published.3–10 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.10 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.5,6 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...
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 hematidrosis with celiac disease in our case might or might not be significant.

To conclude, hematidrosis 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 hematidrosis.

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

<table>
<thead>
<tr>
<th>Test/analyte</th>
<th>Patient result (normal range)</th>
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<tbody>
<tr>
<td>Platelets</td>
<td>241 × 10⁹/L (150–400 × 10⁹/L)</td>
</tr>
<tr>
<td>Prothrombin time</td>
<td>12.9 s (12.3–14.2 s)</td>
</tr>
<tr>
<td>Activated partial thromboplastin time</td>
<td>31.4 s (30.5–40.4 s)</td>
</tr>
<tr>
<td>Factor VIII activity</td>
<td>85% (60–150%)</td>
</tr>
<tr>
<td>Factor IX activity</td>
<td>90% (60–150%)</td>
</tr>
<tr>
<td>von Willebrand factor: Ristocetin cofactor activity</td>
<td>100 IU/dl (50–200 IU/dl)</td>
</tr>
<tr>
<td>von Willebrand factor: Antigen</td>
<td>70 IU/dl (50–150 IU/dl)</td>
</tr>
<tr>
<td>Platelet aggregation (adenosine diphosphate, collagen, arachidonic acid, thrombin, epinephrine, ristocetin) recorded by light transmission</td>
<td>Normal</td>
</tr>
<tr>
<td>PFA-100 (C/Epi cartridge)</td>
<td>112 s (78–199 s)</td>
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</tbody>
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Abbreviations: C/Epi, collagen/epinephrine; PFA-100, platelet function analyser-100.

Conflict of Interest
None.
References