

Case series of bloody sweating; a scary event for families

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Introduction

Bloody sweat is known as hematohidrosis. In this rare clinical phenomenon, patients may develop bloody sweat in conditions of mental or physical stress with undamaged skin [1]. It can be associated with bloody otorrhea, otoerythrosis, bloody tears, or bleeding from the forehead, umbilicus, hand, and leg [2, 3, 4]. The probable etiologies for hematohidrosis include emotional stress, strain, and extreme exertion, fear, vicarious menstruation, systemic diseases, and unknown [1, 5]. Patients might present with bloody sweating mostly from the face, from unbroken skin. It is also necessary to roll out self-injury [5]. The diagnosis is made as witnessed by a medical physician and blood is collected from the intact skin [6, 7]. Treatment with propranolol is effective in patients with hematohidrosis, as well as psychoanalysis, and antianxiety medications are needed in most cases [1, 8]. In this study, we described three cases of hematohidrosis and reviewed the literature. The purpose is to raise awareness of hematologists, pediatricians, and psychiatrists about hematohidrosis.

Case 1

An 8-year-old-girl, Caucasian, presented with bleeding from the undamaged skin of the forearm, leg, and face for 6 months, and these bleeding attacks were repeated almost every 2 weeks (Fig. 1A). The patient had no history of any previous bleeding disorders or coagulation disorders in her family. Her mother without knowing when she was bleeding, tried to control the patient; as such, she did not have self-injury. Her physical examination was normal. She was investigated for bleeding disorders, but all tests, including platelet count (PLT), prothrombin time (PT), partial thromboplastin time (PTT), bleeding time, Factor XIII activity, von Willebrand factor (VWF), fibrinogen level and platelet function test were normal. She was asked to go to the toilet while the interview with her parents was continuing and we saw her bleeding from her face, legs, and abdomen when she came back to her parents (Fig. 1A). Detailed history revealed that most of her bleedings happened when she went to the toilet or when he was feeling lonely. A sample from the bleeding site was sent to the coagulation laboratory, which showed 30% hematocrit and the peripheral blood smears revealed all components of the blood cells.

Propranolol (non-cardio selective beta-adrenergic antagonist) 10 mg, orally, twice per day was started leading partial improvement. Her electrocardiography during treatment with propranolol was normal. As a result, bleeding occurred every two months, rather than almost every 2 weeks. Three months later, she had significant improvement after starting psychotherapy and taking a tricyclic antidepressant (nortriptyline 10 mg, orally, once per day at night) as an antianxiety medication. She is under a close follow-up with no complaint of bleeding events and no adverse events related to her medications were observed.

Case 2

A 12-year-old-girl, Caucasian, was referred to hematologist due to the bleeding episode from ears, eyes, nostrils, and sublingual area almost every 3 weeks without the history of trauma (Fig. 1B). Her physical examination was normal. All coagulation profiles, including PLT, PT, PTT, platelet function test, factor XIII, and VWF were normal. She had no previous history of bleeding disorders in the patient or her family. Although she had no history of anxiety or fear, psychoanalytic consultation was requested and a conflict between the patient and her obsessive mother was detected. With a diagnosis of hematohidrosis, propranolol 10 mg, orally, twice per day was started with minimal response. Her electrocardiography during treatment with propranolol was normal. The bleeding symptoms happened approximately every month instead of almost every 3 weeks. Additionally, as consulted with a pediatric psychiatrist, treatment with antianxiety (nortriptyline 10 mg, orally, once per day at night) along with psychotherapy was associated with partial improvement. She is under a regular follow-up with no new complaint of bleeding events after 7 months and no drug adverse effect.

Case 3

A 14-year-old-girl, Caucasian, who came due to bleeding from the undamaged face (Fig. 1C) when he fought with his sister, who was two years younger, he developed bleeding from the undamaged face and the family was scared and fought with a little sister. Of course, the patient had a history of aggression. The physical examination was normal.

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Fig. 1. Bleeding from hand, foot, and abdomen in an 8-year-old girl after returning from the toilet at an outpatient clinic (A). Bleeding from the ear and forearm in a 12-year-old girl with hematohidrosis (B). Bleeding from undamaged face after a struggle with her sister in a 14-year-old girl (C)

All coagulation tests like in the previous case were done and were found to be normal. According to patient history, physical examination, and the normal coagulation test, hematohidrosis was diagnosed for her. Thus, propranolol was started with a dosage of 20 mg, orally, twice per day, and referred to a psychiatrist that prescribed for her antipsychotic drugs, and psychotherapy with family and the patient was done. Her electrocardiography during treatment with propranolol was normal. Now that one month of patient treatment is over, she has recovered and has not been bleeding for the last month. In the follow-up, she had not been found with any adverse effects related to her medications. Written informed consent was obtained from all the patients and their parents.

Discussion

Hematohidrosis is a rare clinical condition that presents with the bleeding episode from the undamaged skin. It presents with episodes of spontaneous bleeding through intact skin or sweat gland orifices, with an unknown cause [1, 4]. The differential diagnosis of hematohidrosis includes vasculitis, factitious disorder, Munchausen's by proxy, scurvy, pseudo-chromhidrosis, and chromhidrosis [9-13]. In vasculitis syndrome, the patients usually have systemic signs and symptoms. It is important to exactly observe the skin to rule out self-injuries [13]. In scurvy, the patients may present

with bone pain, petechiae, myalgia, easy bruising, swelling, perifollicular hemorrhages, corkscrew hairs, and gum bleeding [14]. Chromhidrosis is characterized by the excretion of colored sweat from apocrine or eccrine sweat glands. In pseudo-chromhidrosis, a much more common disorder, sweat stains are visible after discharge from the sweat gland. Pseudo-chromhidrosis is caused by contact between sweat and dyes, chemicals, or chromogenic bacteria on the skin [10, 11]. Fear and stress are the most common causes of hematohidrosis [15]. The mechanism responsible for bleeding in this disorder includes vasoconstriction of vessels, which supply the sweat glands after anxiety and consequently, in the post-stress phase, vasodilatation occurs, leading to the rupture of the vessels of the sweat glands and the secretion of bloody sweat to the skin [1, 2, 16]. In our report, anxiety and psychological problems were strongly accompanying in both cases, causing bleeding from undamaged skin that had a good response to anti-anxiety medication and psychotherapy. In patients with hematohidrosis, we must be sure to have a history of stress and psychological problems, since it plays an important role in treating such patients. This approach concentrates on alerting physicians, nurses, and health professionals to consider different aspects of psychological problems in such rare bleeding manifestations along with normal coagulation tests. High suspicion to consider hematohidrosis in the pediatric age group presents with bleeding symptoms and normal coagulation profiles is essential to not miss this rare disorder. Psychiatric consultation and initiation of drug therapy with psychotherapy along with propranolol is a good management strategy, as observed in our present cases and previous repeated cases [17]. Although atropine sulfate transdermal patch was used as a treatment option in a study by Biswas et al. [2] the results were almost favorable, but it was gradual. However, in our study, we could not use atropine sulfate transdermal patch due to lack of access, but our patients responded well to propranolol.

Conclusion

As a result, in patients with hematohidrosis, it is better to focus more on psychological problems. Because it seems that stress and the psychological problem has a more prominent role in the etiology of the disease.

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Author's contributions

NS, MS – contributed substantially to the conception and design of the study, analyzed and interpreted the patient data regarding the hematological disease, and also helped in writing the manuscript. AB, MK – followed the patients and helped in writing the manuscript. All the authors – read and approved the final manuscript.

Conflict of interest

The authors declare that they have no competing interests.

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None.

Ethics

The work described in this article has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving humans; EU Directive 2010/63/EU for animal experiments; Uniform requirements for manuscripts submitted to biomedical journals.

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