Case report

A case of haematohidrosis with comorbid attention-deficit hyperactivity disorder and oppositional defiant disorder: challenges to treat

Saqiba Aziz,[1] Shihab Shahriar,[2] Md Arifuzzaman[3]

[1]Consultant Psychiatrist, Medinova Medical Services Limited, Dhaka, Bangladesh; [2]MD Resident of Psychiatry, National Institute of Mental Health (NIMH), Dhaka, Bangladesh; [3]MD Resident of Psychiatry, NIMH, Dhaka, Bangladesh.

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Correspondence
Saqiba Aziz
Mobile: +8801813518532
E-mail: azizsaqiba@gmail.com

Summary

Haematohidrosis is an uncommon disorder of unknown etiology that could be puzzling to physicians. Sufferers of this disorder were often adolescent females who were undergoing some stressful or emotional event; which might be associated with the activation of the sympathetic nervous system. These episodes involved spontaneous bleeding from skin and mucosal membranes without the involvement of trauma. An 11-years-old girl had admitted in a psychiatry hospital with recurrent episodes of bleeding from the eyes, nose and palm for one and a half years. She had been diagnosed with haematohidrosis following through clinical examination and laboratory investigations conducted a few months back prior to admission. However, the treatment appeared to be ineffective, perhaps owing to the patient's comorbid attention deficit hyperactivity disorder (ADHD) and oppositional defiant disorder (ODD). The patient was then treated with combined pharmacological and psychological interventions, thus bringing about an improvement in this challenging case.

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Introduction

Haematohidrosis is a rarely observed clinical disorder that could be puzzling to physicians as it involved the patient experiencing bleeding following splitting of superficial capillaries found of the skin and mucosal surfaces in the absence of trauma or injury. These events have been reported to be precipitated by physical and psychological stress hence the theory that this condition is linked with an overly active sympathetic nervous system.[1] It has also been referred to as hemihidrosis, hematidrosis and hematidros in related medical literature.[2]

In this condition, sometimes, blood from the superficial capillaries from skin surfaces bursts into the surrounding eccrine sweat glands and is excreted along with sweat - thus producing bloody sweat[3]- which is might be why the term haemato-folliculohidrosis was suggested to be describe this phenomenon. Through similar mechanisms, the condition has been linked to systemic diseases and vicarious menstruation.[1] A case of Haematohidrosis has been narrated here who has comorbid attention deficit hyperactivity disorder (ADHD) and oppositional defiant disorder (ODD).

Case summary

An 11-years-old girl was admitted in the National Institute of Mental Health (NIMH), Dhaka, Bangladesh with the complaints of bleeding from the eyes, the nose and the palms for the last one and a half months. The bleeding episodes began after she was admitted a Madrasa. While she performed well academically, she was often restless inside the class and was more interested in playing outside during class hours. Her teachers then subjected her to scolding and hitting physically following which she was seen to bleed suddenly from her eyes, her nose and there was sometimes bloody discharge mixed with the sweat on her palms. The bleeding episodes would last for around 2-3 seconds and seemed to resolve spontaneously. There were no visible injury marks, cuts or other signs of trauma around the injury site. She had no history of joint pain, rashes or diarrhea. With these problems, she was soon removed from her Madrasa and taken to the ophthalmologist who performed several investigations. All the hematological reports including complete blood count, prothrombin time, bleeding time, clotting time, activated partial thromboplastin time (APTT) and thyroid function tests were within normal limits. On the basis of a thorough clinical examination and laboratory investigations, the patient was diagnosed as a case of haematohidrosis and referred to a psychiatrist. The treating physician prescribed her with 10 mg propranolol and 10 mg atomoxetine (as she diagnosed with comorbid ADHD). There were no significant improvements regarding her bleeding episodes, however. Meanwhile she was receiving home

schooling and she began to show demanding behavior and tantrums. The patient would become furious if her demand for toys were not met and she would vindictively beat her mother afterwards. She would also often show defiance towards her mother and father, she would argue with her mother, in particular. Around one week, prior to admission, after an argument with her mother, she went inside her room, leaving the door open and tried to hang herself. She was rescued quickly and later revealed she showed that she impulsively tried to hang herself as a threat to her parents. Thus, she was soon admitted to the NIMH for better management. After admission, she was diagnosed as a case of haematohidrosis with ADHD and ODD. She was treated with sertraline (25 mg once daily) and clonidine (0.3 mg at night); alongside this her mother received parenting training and the patient received sessions of cognitive behavioral therapy under a child psychologist to learn how to cope with anger and stress more effectively. She was also encouraged to take part in daily activities like walking in the play section of the child ward. With these interventions, she began to show improvement during her hospital stay and was ultimately discharged with advice for follow up.

Discussion

Haematohidrosis was a fascinating but perplexing clinical phenomenon in which physicians observe painless, short lived bleeding occurring from the patient's superficial mucosal and skin surfaces. The pathogenesis of phenomenon had been linked to sympathetic overactivity which ultimately leads to rupture of superficial capillaries leads often to extravasation of blood onto sweat glands and superficial surfaces.[4] The diagnosis was made by excluding other possible diagnoses (e.g. Von Willebrand Disease and chromhidrosis) through a series of investigations (e.g. clotting time, bleeding time and platelet count) and detailed clinical examination.

Possible psychosomatic factors were also theorized to be linked to the disorder, seeing as haematohidrosis had in previous reports been often observed in adolescent girls facing stressful life events.[5] While beta blockers had been used in the clinical practice to treat this kind of bleeding[6], for this particular case propranolol was not sufficient to cease bleeding episodes. Perhaps the resolution of the disorder was complicated by the lack of sufficient psychological and pharmacological treatment of the patient's comorbidities, which seemed to act as a perpetuating factor for her haematohidrosis. Along with some medications, proper psychoeducation was given to the patient and patient party; along with proper psychological interventions to teach the patient cope with daily upsets and stressful situations in a more adaptive manner. Improvement of the patient indicated the efficacy of psychopharmacological and psychological interventions for this challenging case.

Conclusion

Haematohidrosis should be kept as an important differential diagnosis in cases where there was bleeding from skin and mucosal surfaces in the absence of visible trauma. As haematohidrosis seemed to be worsen during periods of anxiety and stress, other psychiatric comorbidities should be treated simultaneously. Beta blockers were used in clinical practice to reduce chance of rupture of superficial capillaries caused by excitement but alternatively, alpha 2 adrenergic agonists like clonidine might also work with patients who had similar profile as the one described in this case.

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