The First Case of Hematohidrosis in an Under 10 Years Old Child in Iran: A Case Report

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Abstract

Hematohidrosis is a clinical condition where bleeding occurs through the sweat glands in people older than ten years. However, there was an exception, and there was a child patient under 10 years with unidentified light symptoms. We present the first-reported case of hematohidrosis in a child less than 10 years of age in the Islamic Republic of Iran. The patient was an eighteen-month-old male child who experienced recurrent attacks of hemorrhage provoked by psychogenic stressors. Systematic diagnostic investigations were found to have only iron deficiency anemia because of repeated bleeding episodes. Propranolol treatment resulted in withdrawal of the bleeding attacks. This particular case serves to re-emphasize the need for hematohidrosis to remain in one's mind when taking care of the pediatric age bracket, and that the therapeutic effectiveness of beta-blocker administration applies to its therapy.

Keywords: Hematohidrosis, sweat gland, bleeding, case report

Introduction

Hematohidrosis, the excretion of blood through the sweat glands, is a rare pathology that has been reported mostly in patients above ten years of age. Background reports indicate an association with intense psychological stress, especially reported in combat soldiers and, according to some accounts, in Jesus Christ at the time of crucifixion.[1] The pathophysiologic mechanism beneath is multifaceted, with the vasculature and the sweat glands implicated; profound psychological stress leads to hyperstimulation of the sympathetic nervous system. This causes intense vasoconstriction of the capillary bed that envelops the eccrine sweat glands. Upon alleviation of the stressor, there is a sudden vasodilation that can lead to rupture of the already compromised capillaries. Blood finds its way into the sweat gland ducts and onto the skin surface as bloody sweat. This process is speculated upon by several hypotheses, such as vasculitis of skin vessels that are prompted by sympathetic stimulation and a net-like distribution of capillaries encompassing sweat glands that render the vessels prone to rupture under pressure. This pathophysiologic process accounts for the episodic occurrence of hematohidrosis and the common accompaniment of emotional or physical stimuli[2]

The case is of particular interest as it is a rare case of hematohidrosis in an Iranian child under the age of ten, hence, there is a necessity for increased research into the condition, its etiology, and current therapeutic interventions. The case report in question is an important contribution to the available literature since it is the first documented case to report hematohidrosis in an under-10 patient in Iran. Current research has increasingly been focusing on stress hormone effects, together with sympathetic nervous system activation, on the pathophysiologic mechanisms behind hematohidrosis.[3,4]

Case Report

An 18-month-old male child from Lordegan in the Chaharmahal Bakhtiari province in Iran was referred for assessment as he presented with an ongoing problem in which he experienced repeated bleeding from the external auditory canal. The child's symptoms included periods of bleeding from the external canal over weeks. He referred to ENT specialists to detect source of bleeding but against all the odds, tympanic membrane was normal and there was no ulcer in the external auditory canal. Then referred to Hematologist for bleeding tendency

The standard hematologic investigations done included complete blood count (CBC), bleeding time, prothrombin time (PT, international normalized ratio (INR), partial thromboplastin time (PTT), and liver function tests which all were within normal limits (Plt 170× 109/L, INR 0.7, PTT 24) except for the hemoglobin level (10.2 g/dL) that anemia was based low on for him and low ferritin (8ng/mL). Evaluation for other coagulation factor deficiency like factor XIII done and quantity and function of this factor was in normal range. As the hemorrhage was still severe and lead to decreased level of the ferritin, the patient received iron supplement (ferrous sulfate 5 mg/kg/day) and was followed up with CBC and physical examination on several occasions.

Then, one of the incidents that was witnessed was when the child was trying to have his hair cut. The child started crying and sweating on the head especially the forehead, which also had sores. Over time the person started having bleeding episodes in the axillary and inguinal regions.

Upon realizing that the child has a health issue, the family reported instances of conflict and distress in home settings. However, the child showed no signs of stress which include hematohidrosis given the stressful situation the child was going through. The rest of the family also shared that the child has certain phobias that make the child manifest symptoms when exposed to certain triggers. For example, when the child wanted to go to the park and hear the barking of the neighbor's dog, the child developed fear and started crying and bleeding [Figure1].

Figure 1: Bleeding when the child is scared and crying.

The person in question lived in Lordegan, in the Chaharmahal Bakhtiari a province of Iran. When the child was two years old, the family came to us. The physician noticed that when the patient transferred from his living lesion to Shiraz, bleeding stopped.

This realization made the family move to Shiraz to reduce the chances of having hemorrhages. However, every time the person visited Lordegan, the symptoms of hematohidrosis worsened, thus demonstrating the crucial effect of the fear triggers on the person's health.

Stress plays a significant role in the development of this disease and therefore needs to be given team a lot of attention prescribed in propranolol in this case. After 1mg/kg/day of it, patient responded to this dose of medication. After 3 months, the patient's mother refused to take the medication for him, that caused repeat the bleeding. This was resolved by retake the medication. Then, after a few months, the patient was placed under the supervision of the psychiatric team and was controlled by continuing the process of propranolol and psychological treatments.

Discussion

Hematohidrosis is an unusual clinical condition wherein blood is excreted through sweat glands. Though pathophysiological mechanisms responsible for this condition are not fully known, changes in vasomotor tone resulting from stress have been thought to result in rupture of capillaries in sweat glands. Psychological stressors, functioning of the autonomic nervous system, and underlying vascular malformations have been cited as possible contributory factors for this condition[5,6]

Most applicable to the case is the function of environmental factors. The family noted a reduction in bleeding incidents when the child for trial was relocated from Lordegan to Shiraz, which indicated that environmental stressors could have been exacerbating the condition.[7] The observation once more indicates the necessity for an understanding of the environmental factor and the institution of stress breakers for hematohidrosis control.

In the case being considered, an eighteen-month-old male presented with recurrent bleeding from the external auditory canal, which further detailed hematological evaluation diagnosed as hematohidrosis. While initial assessments indicated the development of iron deficiency anemia, the association of the hemorrhagic incidents with stress-provoking situations, including haircuts and exposure to phobias (i.e., barking dogs), indicates a profound psychological factor.[8]

Pharmacological intervention using beta-adrenergic antagonists like propranolol has yielded mixed results in treating hematohidrosis. In our study, propranolol administration led to resolution of hemorrhagic attacks within three months, thus affirming previous observations on the efficacy of beta-blockers in stress-related disorders.[9] Beta-blockers have been proposed to suppress stress reactions by dampening sympathetic nervous system function and restoring normal vasomotor reactions.

Beta-blockers, a class of medications that competitively antagonize β-adrenergic receptors (primarily β_1 and β_2 subtypes) to inhibit the effects of catecholamines, such as adrenaline and noradrenaline, have been proposed to suppress maladaptive stress reactions by dampening the hyperactivity of the sympathetic nervous system (SNS).[9,10] The SNS, the body's primary mediator of the "fight-or-flight" response, becomes overactive during chronic stress, leading to excessive cardiovascular and metabolic activation. Beta-blockers counteract this by reducing heart rate, myocardial contractility, and blood pressure through β_1-receptor blockade in cardiac tissue, while β_2-receptor inhibition in vascular smooth muscle decreases peripheral vascular resistance. Furthermore, beta-blockers restore normal vasomotor tone by stabilizing dysregulated vascular responses caused by SNS overactivation.[10,11]We initiate 1 mg/kg/day of propranolol divided in 2 doses for him with education to his parents to monitor heart rate by repeated checking of his pulse. Our patient had no history of asthma or bradycardia and could tolerate this dose of it. He did not show any side effects of propranolol like Fatigue, dizziness, hypoglycemia, and cold extremities.

Conclusion

This case report records the first reported case of hematohidrosis in a patient younger than ten years in Iran. The successful management of hemorrhagic attacks with propranolol and the identification of environmental stressors highlight the importance of a multidisciplinary approach in the management of this unusual condition. Future research

should endeavor to elucidate the precise pathophysiology of hematohidrosis and determine ideal therapeutic protocols, both pharmacologic and psychosocial therapy.

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