

# Hematohidrosis: A Rare and Mysterious Case

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## ABSTRACT

**Background-** Hematohidrosis is a rare condition characterized by the spontaneous discharge of blood or blood components through intact skin. This case report presents a case of a 12-year-old Indian girl who presented with recurrent spontaneous bleeding while walking mixed with sweat from intact skin, diagnosed as hematohidrosis, to understand the condition better.

**Methods-** The study presents a case study of a 12-year-old female patient with hematohidrosis and discusses the treatment approach using propranolol. The patient was found to be bleeding from intact skin of the tendoachilles region right ankle on walking. The patient had a history of trauma 15 days back, but there is no abrasion or discoloration associated with bleeding or due to infection and was alert and had no abnormality in general examination. The secretion from the area was collected and processed for the benzidine test and microscopic examination. The bleeding was cleaned and the area was examined for trauma or self-inflicted wounds.

**Results-** The present study showed that blood secretion was spontaneous and positive, with no abnormalities. Biochemical tests were normal, and ultrasonography was normal. Vitamin C and propranolol were prescribed, and the patient did not have a recurrence of the bleeding episode until two months during academic exams. Anxiety and stress may have exacerbated the bleeding. A repeat dose was advised, and the patient did not report any bleeding complaints for six months.

**Conclusion-** Propranolol was used as an efficient beta blocker against the condition, and stress was declared as one of the contributing factors for hematohidrosis.

**Key-words:** Blood sweating, Hematohidrosis, Propranolol, Recurrent hematohidrosis, Psychological stressors

## INTRODUCTION

Hematohidrosis or Hematidrosis is a rare condition with characterized recurrent episodes of spontaneous bleeding in normal skin and intact mucosa. A related instance is mentioned in Leonardo da Vinci's description of a soldier under extreme stress. Also, this phenomenon has been proposed as an explanation for Jesus Christ's crucifixion in the garden of Gethsemane and the claims associated with stigmata<sup>[1,2]</sup>. The possible explanations for hematohidrosis can be systemic disorders, bleeding

disorders, vicarious menstruation, excess exertion, and psychological stressors<sup>[3]</sup>. The exudation of blood is caused by the rupture of capillary blood vessels that nourish sweat glands due to stress<sup>[4]</sup>. Previous literature suggests such bleeding from skin (hematohidrosis), eyes (hemolacria), and ears (blood otorrhea)<sup>[2,5,6]</sup>. On the other hand, Holoubek et al. [4] reported one of the first kinds of hematohidrosis in the genital and perineal area. The exact cause of hematohidrosis is unknown, but it has been associated with anxiety disorders. Laboratory investigations, including blood workup and systemic examination, are usually unremarkable. Hematohidrosis can also be associated with systemic diseases and extreme exertion<sup>[7,8]</sup>. It may sometimes be triggered by specific events, such as school examinations or quarantine periods<sup>[9]</sup>. Treatment with propranolol has shown successful outcomes in some cases. Beta-blockers, such as propranolol, are useful in managing

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hematohidrosis<sup>[10]</sup>. However, no definitive treatment is available for this condition<sup>[11]</sup>. Psychological anxiety has been identified as a predisposing factor for hematohidrosis<sup>[12]</sup>. The condition can be effectively managed if the underlying cause is correctly identified and addressed. Healthcare professionals need to provide psychological counseling and reassurance to both the patient and their parents, as hematohidrosis is a benign condition<sup>[13]</sup>. Although specific guidelines for treatment are not well established, hematohidrosis can be successfully treated with a combination of pharmacotherapy and non-pharmaceutical interventions. We now present a case of a 12-year-old Indian girl who presented with recurrent spontaneous bleeding while walking mixed with sweat from intact skin, diagnosed as hematohidrosis, to understand the condition better.

#### CASE PRESENTATION

A 12 years old female child resident of Raipur, Chhattisgarh, India, was presented to the Dept of General Medicine, Shri Balaji Institute of Medical Science, Raipur, on 25 July 2023 with complaints of spontaneous bleeding from intact skin of the tendoachilles region right ankle on walking. The bleeding was noted to be spontaneous and recurring. The patient had a history of trauma 15 days back, but there is no abrasion or discoloration associated with bleeding or any other bleeding sites such as gums/ oral mucosa/ sub conjunctiva/ epistaxis/ hematoma; scurvy and other bleeding dyscrasias were ruled out. Bleeding was found to be not associated with menstruation. Also, no family history of any bleeding tendency was noted. The patient had complaints for 15 days that occurred while walking (Fig. 1). She gives a history of recurrent bleeding on walking multiple times a day. She advised me to follow in OPD.

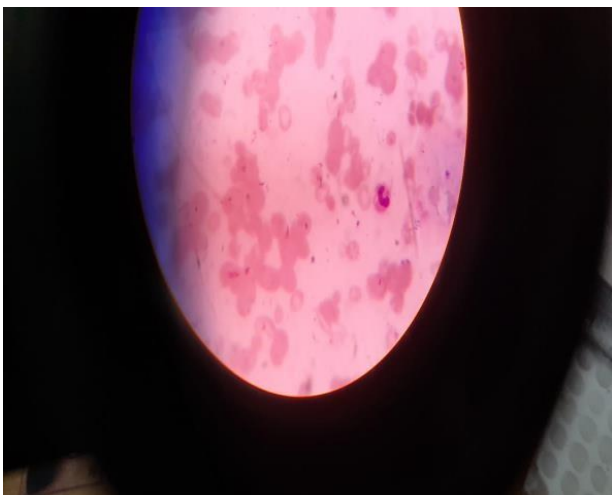
The patient was alert and no abnormality was noted on general examination. She was oriented, comprehended, and communicated well with people nearby. No psychotic symptoms were elicited and her intelligence was observed to be within normal limits. On detailed examination, no physical or gynaecological abnormality was noted. The secretion from the area was collected and processed for benzidine test and microscopic examination (Fig. 2). The bleeding was cleaned and the area was examined for trauma or self-inflicted wounds.

Absence of injury was noted and the skin over the tendoachilles region right ankle appeared normal. The absence of systemic disease and allergy towards drugs or food was noted. Later, in the OPD another episode of bleeding was observed and was witnessed by three physicians. The blood secretion was found to be spontaneous for one to two minutes. On biochemical examinations, the secretion from the tendoachilles region right ankle was positive for blood components with no other abnormalities. Complete blood count, bleeding and clotting time, prothrombin time, active partial thrombin time, liver function tests, and renal function tests are within normal limits. Ultrasonography of the abdomen and pelvis was found to be normal. The secretion was confirmed to be blood by benzidine test and microscopic examination also revealed blood elements in the sample.



**Fig. 1:** (A) Bleeding starts on walking (B) Normal skin when not walking

As informed by the patient and her family, symptoms usually appear when she walks, and she is afraid or stressed, particularly in the morning. Also, in the hospital, while she walks and expressed fear and stress regarding her academic performance. The patient was prescribed vitamin C daily and propranolol 10 mg twice daily for ten days and tapered for 1 month. No recurrence of the bleeding episode was noted until two months during her academic exams. The possibility of exacerbation associated with anxiety and stress was noted. Propranolol repeat dose was advised and the patient did not report any such bleeding complaints for six months.



**Fig. 2:** Smear prepared from the oozing blood

## DISCUSSION

Hematidrosis is an extremely rare and enigmatic disorder characterised by recurrent episodes of spontaneous bleeding from intact skin, which is usually self-limiting. The etiopathogenesis for the condition, as proposed by a few authors, includes post-systemic disease, excessive exertion or stress, vicarious menstruation psychogenic and unknown [2,14-16]. In most of the cases, acute stress and anxiety were the precipitating factors. In our case, the patient was advised vitamin C owing to its probable role in sympathetic activity and anxiety. The phenomenon was found to be isolated and was not found to be associated with general health conditions.

Duan *et al.* [17] reported hematohidrosis associated with primary thrombocytopenic purpura. Diagnosis was made by exclusion and clinical findings of peripheral blood exudates of intact skin. In our study, no such abnormality was found. There is a lack of knowledge

about the pathology of the condition. However, some studies have suggested increased vascular pressure leading to the passage of blood cells through the duct of sweat glands, vasculitis of dermal vessels and exacerbated sympathetic activation leading to peripheral glandular vessel constriction and subsequent expansion, allowing the passage of blood content into ducts [15].

Hematohidrosis should be differentiated from chromatidrosis, meaning coloured sweat due to coloured apocrine secretion and also from other haemorrhagic disorders, including platelet abnormality, coagulation or vasculature abnormality, which usually is found to be associated with petechiae, purpura, mucosal bleeding or hematoma. Differential diagnosis for the disorder must include chromhidrosis, factitious dermatitis, vicarious menstruation, vasculitis, and platelet and coagulation disorders. Lab investigations are of utmost importance.

To date there, no specific treatment for the disease has been identified. However, Manolukul *et al.* [18] used lorazepam as anxiolytic and significant results were obtained. Badry *et al.* [19] treated their case of a 16-year-old female patient complaining of recurrent attacks of right-sided bleeding with propranolol 10 mg twice daily. After 2 weeks of the treatment, the episodes were found to have decreased. This case and treatment methodology used to decrease the episode is similar to our case and results [20]. The sympathetic system has a certain role in the recurrent episodes of hematohidrosis, which is usually common during emotional or physical stress, as in our case. The patients were found to be relieved, and recurrence of hematohidrosis decreased on administration of beta blockers, similar to our study. Bhattacharya *et al.* [21] noted diazepam's ineffectiveness and reported effective results with propranolol, thus recommending propranolol and supporting the treatment used in our study.

## CONCLUSIONS

We treated our patient with propranolol 10 mg twice daily based on its similar uses in the literature and its effective observations. This led to a significant observation and remission was achieved to date. In conclusion, stress is noted to be a contributory factor for the progression of the abnormality. The phenomenon is noted as a rare condition, and similar and associated case studies are advised for a better understanding of the disease and to devise a treatment protocol.

## CONTRIBUTION OF AUTHORS

**Research concept-** Naresh Verma

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