



Understanding Hematohidrosis: A Rare Phenomenon in Infancy - Case Report

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Abstract

Hematohidrosis, a rare condition characterized by the excretion of blood through the sweat glands, shows diagnostic challenges due to its scarcity. A 2-month-old male infant presented with recurrent bleeding from his nose, eyes, and ears, which appeared to be sweating blood. Clinical examinations, including imaging and laboratory investigations, were unremarkable. The diagnosis was made by exclusion criteria and confirmed with the administration of anxiolytics (lorazepam) and propranolol, which significantly reduced the bleeding episodes. The patient remained asymptomatic after discontinuation of propranolol. This case underscores the importance of clinical vigilance and comprehensive evaluation in diagnosing and managing hematohidrosis, particularly in infants, where prompt intervention can alleviate symptoms and prevent complications.

Keywords: Hematohidrosis, infant, bleeding, anxiolytics, beta-blockers

Introduction

Hematohidrosis is a very rare condition in which a person sweats blood. Few cases of hematohidrosis have been reported in the literature. This condition, also known as hematidrosis or hemidrosis, occurs when capillary blood vessels that feed the sweat glands rupture, causing blood to exude. It typically occurs under extreme physical or emotional stress (Freddrick, n.d., Badry et al., 2020).

Severe mental anxiety activates the sympathetic nervous system, triggering the stress-fight-or-flight reaction to such an extent that it causes hemorrhage of the vessels supplying the sweat glands, leading to blood entering the sweat ducts (Jerajani, Jaju, Phiske, & Lade, 2009).

Episodes of hematohidrosis may be preceded by intense headache and abdominal pain and are usually self-limiting. The secreted fluid can vary in appearance; in some cases, it is more dilute and blood-tinged, while in others, it is darker and bright red, resembling blood. The effects on the body include weakness and mild to moderate dehydration due to severe anxiety and the loss of both blood and sweat (Biswas, Surana, De, & Nag, 2013).

The exact etiology of hematohidrosis remains unknown. On February 23, 2019, a two-month-old male infant was brought to my private clinic by his parents, with the chief complaint of bleeding from his nose, eyes, and ears (Zhang et al., 2004).

The condition started at the age of one month as frequent episodes of bleeding from the ear (blood otorrhea), eyes (hemolacria), and nose (epistaxis). The episodes occurred every 2-3 days, initiated by

Significance | This case highlights the diagnostic challenges and successful management of hematohidrosis in infants, shedding light on effective treatment strategies.

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crying followed by bleeding, and then the infant would sleep. During each episode, the patient was admitted to the hospital and received IV fluids and vitamin K. After about an hour, the bleeding stopped, and the patient was discharged without a history of receiving blood transfusions, as the parents reported that the packed cell volume (PCV) did not decrease below 40%. The episodes did not occur during sleep.

During this period, the parents consulted many doctors, but none could reach a diagnosis. The patient remained active and fed well during and between episodes. The infant was delivered by cesarean section to a G3P3A0 mother and was postnatally active without complications. The umbilical stump fell off after 10 days without bleeding. The infant was on bottle feeding (hydrolyzed formula prescribed by one of the doctors). The parents had separated about six months prior. There was no family history of bleeding tendencies or systemic diseases, nor was there evidence of trauma.

On general examination, the infant was very active, had good muscle tone, and systemic examination revealed nothing significant—no lymphadenopathy, organomegaly, or petechiae. However, there was oozing of blood from the ears, eyes, and nose (see Figure 1, 2). After wiping, the blood disappeared without signs of trauma at the bleeding sites, reappearing within seconds, giving the impression it was sweating rather than bleeding.

The patient was referred to an ENT specialist, who found no sign of injury or ear disease. All investigations, including CBC, liver function tests, renal function tests, and tests for bleeding tendencies, were normal (bleeding time 4 minutes, prothrombin time 15 seconds, activated partial thromboplastin time 30 seconds, platelet count 200,000). Platelet function tests, von Willebrand factor, and all coagulation factors (including factor XIII) were normal. Brain ultrasound and CT scans were also normal.

The family was reassured, the infant's formula was changed to standard formula, and the patient was prescribed oral lorazepam (0.05 mg/kg/day) and propranolol (0.6 mg/kg/day) for two months. During this period, the attacks occurred only twice and with less severity, confirming the diagnosis of hematomhidrosis. Lorazepam was then tapered off, and the patient remained on propranolol. At the age of five months, the parents consulted about circumcision, which was performed without complications (see Figure 3). At seven months, propranolol was discontinued, and the child has continued to live without any problems.

The diagnosis of hematomhidrosis is made clinically by exclusion and confirmed by the reduction and eventual cessation of bleeding episodes with the use of anxiolytics and propranolol.

Discussion on hematomhidrosis Diagnosis

Hematomhidrosis, also known as hematidrosis, hemidrosis, or hematomfolliculohidrosis, is an enigmatic disorder characterized by recurrent episodes of self-limiting bleeding from intact skin. It can

occur on any part of the body and at several points simultaneously (Patil et al., 2015). Despite its extreme rarity and the lack of a clear scientific explanation to support its existence, hematidrosis is real and has been reported for many centuries worldwide (Caproni et al., 2017). Diagnosis of hematidrosis can only be made if the following criteria are met: i) recurrent, spontaneous, painless, and self-limited oozing of bloody discharge is witnessed and confirmed by health professionals, ii) the usual blood components are found in biochemistry studies of the discharge, and iii) the site of bleeding is intact with no abrasion, telangiectasia, or purpura, and after wiping the area, there is no evidence of oozing. All these criteria must be met to rule out organic bleeding disorders, self-inflicted bleeding, factitious disorder by proxy, and chromhidrosis (Holoubek et al., 2017).

The etiology and pathogenesis of hematomhidrosis remain unclear. Although experts consider extreme physical or emotional stress to be the main cause, it may occur without any preceding stressful situation (Raj et al., 2013). In our case, an infant was brought to a clinic with recurrent bleeding episodes from the nose, eyes, and ears, starting at one month of age and triggered by crying. Extensive medical evaluations, including blood tests and imaging studies, yielded normal results, making a definitive diagnosis elusive initially.

There are a number of theories regarding the explanation of hematomhidrosis. The most commonly proffered explanation relates to intensified sympathetic activation due to extreme physical or mental stress. The "fight or flight response" invoked by sympathetic activation leads to the constriction of capillary vessels feeding the sweat glands (Biswas et al., 2013). When the anxiety subsides, the blood vessels dilate to the point of rupture, leading to the passage of blood through the ducts of the nearby sweat glands, presenting as droplets of blood mixed with sweat on the intact skin surface or mucosa in almost any part of the body. Such manifestations may occur at several points simultaneously (Gupta et al., 2016).

Dermal vasculitis is also concluded as a pathological basis for hematomhidrosis. Stromal weakness due to defects in the dermis is another theory to explain the occurrence of hematomhidrosis. According to this theory, the communication between these defects and vascular spaces in the dermis may lead to the establishment of dilated blood centers. Whenever the positive pressure inside vascular spaces exceeds a certain level, blood will exude via follicular canals or directly onto the skin surface (Duan et al., 2014). Subsequently, they will collapse and leave no scar.

This phenomenon, which acts like a balloon, will wax and wane, thus explaining why the bleeding is intermittent and self-limiting (Freddo et al., 2012). The bleeding is intermittent because the vascular spaces disappear after exuding their content but reoccur once the blood flow is reestablished. An immediate biopsy is important for a definite diagnosis. Biopsy during a symptom-free



Figure 1. Oozing of blood from the eyes and nose.



Figure 2. Oozing of blood from the ear and cheek.



Figure 3. Five years old child after treatment without complications.

period does not reveal any blood-filled vascular spaces, intradermal bleeding, or abnormality in hair follicles and sebaceous or sweat glands (Murgia et al., 2013). The term “hematofolliculohidrosis” is proposed as it appears along with sweat-like fluid, and the blood pushes via the follicular canals (Pardo et al., 2012).

In some cases, there is no convincing specific therapy available for rare condition of hematidrosis, though there are reports of good response to various drugs such as anxiolytics, especially in cases triggered by extreme stress. There are some reports of successful use of propranolol (Beccuti et al., 2012). Atropine sulfate transdermal patches have also been used successfully. In addition, in a case with simultaneous epileptic seizure and hematidrosis, the symptoms of both were successfully resolved following the administration of the anti-epileptic drug, oxcarbamazepine (Salvi et al., 2015). Hematohidrosis rarely causes serious side effects, though some people experience dehydration and anxiety. Doctors may give additional medication to treat these symptoms. Psychological counseling can also help if a person with hematidrosis has depression and anxiety. In the case of our patient, beta-blocker, sertraline, supportive psychotherapy, and relaxation therapy diminished the frequency of episodes.

Conclusion

Hematohidrosis is an extremely rare medical condition, with only a handful of cases reported worldwide. Although the bleeding associated with hematohidrosis can appear alarming, it generally does not adversely affect the overall health of the patient. The use of anxiolytics and beta-blockers has been demonstrated to be an effective treatment, helping to reduce the frequency and severity of episodes. This management approach offers significant relief to patients and supports their long-term well-being.

Author contributions

K.M.A.D. conducted the study, wrote, reviewed, and edited the paper.

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Competing financial interests

The authors have no conflict of interest.

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