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Paediatrics

HEMATHIDROSIS – A RARE CASES OF MALE AND FEMALE CHILD WHO SWEAT BLOOD

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ABSTRACT Hemathidrosis is a very rare condition of eccrine sweat disorder chracterized by one or more episodes of spontaneous bloody sweating from non traumatized skin .A male child who presented to us is reported . In the recent past total of 25 cases reported out of which 21 were women and median age was 13 years and majority 62% from asia mainly india . Main locations were face including forehead, eyes and ears. Prodromal symptoms in 36% ,Possible triggering factors in 86 % identified .Herein we describe a case of hemathidrosis of 12 year old male child presented to our department ,confirmed by testing the secretions for blood components and by excluding all other bleeding disorders and who responded to proponolol therapy

KEYWORDS :

INTRODUCTION

Hemathidrosis is a rare condition in which a human being sweats blood [1].The word hematofolliculohidrois was proposed by manonukul et.al because it appears sweat like fluid and the blood excluded via the follicular canals[3] . various causative factors have been suggested by holoubek and holoubek like component of systemic disease , vicarious menstruation ,excessive exertion, psychogenic purpura and unknown causes [2]. Acute fever and intense mental contemplation are the most frequent cause . Treatment of underlying etiology partly helps in remission of symptoms however even in those cases with systemic etiology psychological factor acts as a precipitating factor for bleeding .There are case reports of bleeding from skin eyes and ears called hemathidrosis hemolacria and blood otorrhea respectively

CASE REPORT

- A 12 Years male child was admitted to the hospital as a case of unusual painless bleeding from the left temple , ears and nose .the consistency of bleeding was little thinner than blood and stopped as soon as the site was wiped leaving behind no oozing site
- Two episodes of bleeding episodes seen during hospitalization and also presence of recorded video evidence of bleeding from nose and ears in home without any reasonable cause such as trauma etc
- The episodes of bleeding started 1 yr back and at the start about 3 episodes per day to approximately 20 episodes per month were observed by care takers until child presented to us
- on thorough interaction with the parents , friends and care takers we observed that the episode occurs when he hears the word **HOSTEL** when he was in home and **Name of the lecturer** who he was afraid of when he was in hostel
- The patient doesnot give any history of aura and pain during episodes and any ingestion of anticoagulants and other drugs ,topical application of medication or exposure to dyes there is no history of any chronic medical condition or similar history in family
- The child was active and no abnormality was detected on general examination On palpation of area no tenderness observed and blood could not be extruded on pressure
- Systemic examination was normal
- We observed 2 episodes of bleeding[clinical figure1 and 2] during
- hospital stay and on examination the secretion was bright red less viscous than blood .there was no sign of injury over temple region or trauama to ear
- The secretion was immediately collected for examination and later mopped up after which no bleeding occurs

LAB INVESTIGATIONS

CBC
Hemoglobin - 11.8gm/dl
RBC - 4.21 million/cubicmm
WBC - 6100 /mm3
PLATELET COUNT - 2,51000/mm3
BLEEDING TIME - 1 min 30 sec
CLOTTING TIME - 3 min 15 sec

COAGUALATION PROFILE

THROMBIN TIME - 19 sec [control 20 sec]
FIBRINOGEN - 207 mg/dl
IVYS BLEEDING TIME - 1min 30 sec
CLOT SOLUBILITY TEST - normal
PROTHROMBIN TIME- 15 sec [control 13 .5 sec]
ACTIVATED PARTIAL THROMBOPLASTIN TIME - 31 sec [control 30 sec]
FACTOR VIII ACTIVITY - 121% [normal 60-150]
VWF:AG LEVELS - 77% [normal 50-150]]
ANA studies - NORMAL
PERIPHERAL SMEAR- NORMAL STUDY

PLATELET AGGREGOMETRY WITH 4 REAGENTS

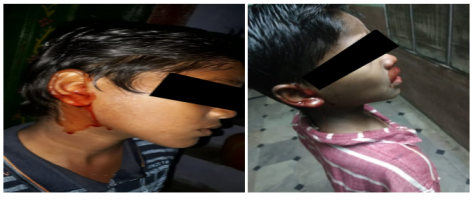
ADENOSINE DIPHOSPHATE - 61% [61-82]
COLLAGEN - 79 % [62-82]
RISTOCETIN - 87% [66-97]
EPINEPHRINE - 80% [62-82]

MICROSCOPIC EXAMINATION

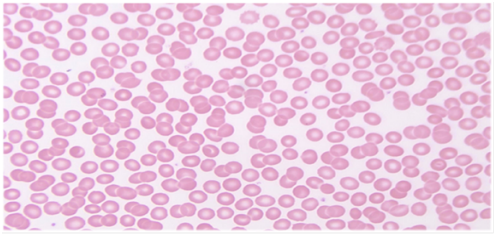
Revealed plenty of RBCS[photograph 3]

The child was managed with 1mg/kg/day of propronolol for past two months and adviced psychiatrist counselling sessions monthly in view of acute fear and anxiety symptoms as triggering factors as mentioned above . Decrease in episodes of bleeding seen after intervention

Clinical photograph 1 and 2

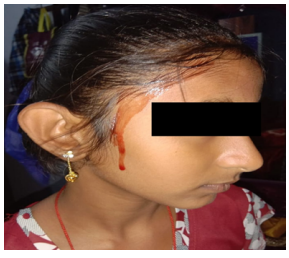


Clinical photograph 3



CASE REPORT 2

- A 12 yrs old female child was admitted in the hospital in view of painless blood like material coming from scalp region and right temple region and occasionally from nostrils and ear canal and stopped as soon as site is wiped leaving behind no oozing site
- Two episodes of bleeding observed at the time of admission and consistency of material was thinner than blood without any trauma and bleeding from other sites
- The episodes started 3 months back and with more than 5 episodes per day from above mentioned sites
- Parents consulted several local doctors and referred to GGH kurnool in view of any bleeding disorder
- On interaction with parents history of unusal frightening dreams experienced by patient from the past 5 months
- Episodes are not coinciding with dreams but occuring after the event in night without any triggering factor in the day times as observed by parents at home and at hospital
- The day on which we observed episode is preceded by some fearful dreams in the night
- All lab investigations CBC, coagulation profile
- Factor viii levels , peripheral smear
- PLATELET AGGREGOMETRY
- BLEEDING TIME , CLOTTING TIME
- THROMBIN TIME, FIBRINOGEN ARE NORMAL
- Microscopic examination reveals plenty of rbc
- The child was managed with propanolol 1mg/kg/day
- Psychiatric counsellings advised
- Decrease in episodes of bleeding seen after follow up



DISCUSSION

Hematohidrosis is an enigmatic disorder characterized by recurrent episodes of self-limited bleeding from intact skin.[4] It is also known as hematidrosis and hemidrosis. It is a well-recognized diagnosis according to the International Classification of Diseases (ICD9-CM 705-89). It is an extremely rare condition, in which capillary blood vessels that feed the sweat glands rupture causing them to exude blood. It can occur at any part of the body and at several points simultaneously and the amount of bleeding is usually small. Most common site is face with some reports of bleeding from eyes and ears also leading to bloody tears and blood otorrhea. Other sites include trunk, limbs, and rarely palms, soles, and mucosa. Exact etiology is not known though various factors are attributed from bleeding disorders to psychogenic and unknown causes.[2]

Hysterical mechanisms and psychosomatic disorders are also believed to induce bleeding.[3] Acute fear ,emotional stress intense mental contemplatation is the most common cause. In our case,acute fear and emotional stress was a trigger for bleeding.Etiopathogenesis of hematohidrosis has been explored in some of the previous studies with no clear conclusive evidence. It has been proposed by Dr.Frederick Zugibe that multiple bloods vessels present in a net-like form around the sweat glands constrict under the pressure of stress and as the anxiety passes out vessels dilate to point of rupture and blood goes into glands and pushed to the surface.[5] Severe anxiety activates sympathetic system to invoke the stress-fight or flight reaction to such a degree as to cause hemorrhage of the vessels supplying sweat glands into their ducts

In our case, the no pain and tenderness after each episode of bleeding noted which has been seen in few cases in literature. Manonukul et al. have proposed that there may be some dermal defects that communicate with dermal vascular spaces and eventually dilate as blood comes in and later exude this blood through follicular canals to collapse without leaving any scar. This phenomenon acts like a balloon, waxing, and waning, thus causing intermittent and self-limited episodes of bleeding. Immediate biopsy soon after an episode of bleeding may reveal the dermal defects as blood filled spaces.[3] A study by Zhang et al.[6] revealed some intradermal bleeding and obstructed capillaries with no abnormality in sweat glands, hair follicles, and sebaceous glands. They concluded that a distinctive vasculitis might be the pathological basis for hematohidrosis.[7] Diagnosis of hematohidrosis is made on the presence of bloody discharge without any obvious cause,through intact skin, witnessed and confirmed by health professional and presence of blood components on testing the bloody discharge.[8] This condition is usually self-limiting in nature with a good prognosis.

Currently, there is no convincing specific therapy available for this rare condition though there are reports of good response to various drugs such as anxiolytics, especially in cases triggered by extreme stress.[3] Various authors have reported good response to propranolol given in a dose of 1 mg/kg/day in two divided doses.[4,7,9] Successful use of beta-blockers supports the role of sympathetic nerve activity in the pathogenesis of this disease. Recently ,Biswas et al. have reported a case successfully treated with atropine transdermal patch.[10]We report this cases for its rarity and the bleeding from external auditory canal , temple and nasal mucosa which is rarely seen in literature and also for its clinical response to propranolol.

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