Original Research Article

Spontaneous ear nose throat bleed: hematohidrosis an unknown entity
series of eight cases

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ABSTRACT

Background: Spontaneous bleed from ear, nose, throat is a rare condition called as hematohidrosis, which occurs under condition of extreme physical or emotional stress. This condition manifests as spontaneous painless bleeding through unbroken skin in any part of the body. It is self-limiting in nature. This condition is relatively unaware among otorhinolaryngologists, the diagnosis of which is made by presence of bloody discharge without any obvious cause through intact skin, witnessed and confirmed by a health professional and the presence of blood components on biochemical analysis.

Methods: Retrospective analysis of series of eight cases.

Results: All eight cases had normal haematological parameters and were under emotional stress. Two cases manifested with oral bleed.

Conclusions: Hematohidrosis is relatively unknown condition to the otorhinolaryngologists and the cases are occurring sporadically which goes unnoticed. This condition occurs in severe stress which may also lead to ear, nose, oral, gastrointestinal bleeds or in combination from any of the sites with a relation of severity of stress level with quantity and site of the bleeding episodes. The condition can be managed effectively if the condition is recognized after ruling out bleeding diathesis. If awareness is created about this clinical condition which is relatively unknown entity, it will lead to further insight on evaluation and management of this rare entity probably an exocrine disorder.

Keywords: Spontaneous bleed, ENT, Hematohidrosis

INTRODUCTION

Spontaneous bleed from ear, nose or throat with no pathological cause under severe physical or emotional stress is encountered by ENT clinician. This is a rare clinical condition called as Haematohidrosis. This is a spontaneous painless bleeding through unbroken skin in any part of the body. Blood sweating is called haematohidrosis, while true bleeding occurs in bleeding disorders. Migliorini described a case of hematohidrosis, which is otorrhea with otoerythrosis.

The term “hematofolliculohidrosis” was proposed by Manonukul et al, because it appeared along with sweat-like fluid and the blood exuded via the follicular canals. Holoubek suggested components of systemic disease, vicarious menstruation, excessive exertion, psychogenic, psychogenic purpura and unknown cause. Acute fear and intense mental contemplation are the most frequent causes.

The diagnosis of hematohidrosis is made on the presence of bloody discharge without any obvious cause through intact skin, witnessed and confirmed by medical health professional/doctor and the presence of blood components on biochemistry studies of the discharge. Hematohidrosis is a well-recognised diagnosis; (ICD-9-CM:705-89).
METHODS

Retrospective analysis was done.

Inclusion criteria

- Spontaneous bleed from ear, nose, throat, or a combination from these sites.
- Age group - Above 12 years.

Exclusion criteria

- Underlying bleeding disorders, local ENT pathology, physical abuse, allergy, hypertension.

Workup

All patients undergo ear, nose, throat examination.

- Endoscopy: Diagnostic nasal endoscopy, video laryngoscopy, upper gastrointestinal tract endoscopy (for oral bleed manifestation).
- Hematological evaluation and opinion for all cases were obtained.

Cases

Figure 1: Right ear bleed one episode.

Figure 2: Both ears bleed one episode.

Figure 3: Right ear bleed two episodes.

Figure 4: Right external auditory canal bleed one episode.

Figure 5: Right external auditory canal bleed two episodes.

Figure 6: Nasal bleed from philtrum one episode.
bloody secretion through intact skin or sweat gland orifices without an unknown cause. Hysteric and psychosomatic disorders are also believed to induce bleeding. Psychogenic purpura is supposed to be caused by hypersensitivity to the patient’s own blood or autoerythrocyte sensitization and is characterized by repeated crops of ecchymoses, gastrointestinal bleedings and hematuria. Another type of bleeding through skin is psychogenic stigmata, a term used to signify areas of scar, open wounds or through unbroken skin. Patients belonging to this group were found to be neurotic. Copeland reported a patient who developed bleedings from her old scars whenever she had serious anxiety. The etiopathogenesis proposed by Dr. Frederick Zugibe is that multiple blood vessels around the sweat glands arranged in net-like form. It is believed that under pressure of great stress the vessels contract. Subsequently as the anxiety passes the blood vessels dilate to the point of rupture. The blood at this point goes into the sweat glands, which pushes the blood to the surface and manifests as droplets of blood mixed with sweat. The extravasated blood has identical cell components as that of peripheral blood. The severe mental anxiety activates the sympathetic nervous system to invoke the stress-fight or flight reaction to such a degree as to cause haemorrhage of the vessels supplying the sweat glands into the ducts of sweat glands. Effect on the body is weakness and mild to moderate dehydration from the severe anxiety and both blood and sweat loss. Skin histopathological study by Zhang et al, revealed some intradermal bleeding and obstructed capillaries. No abnormality was found in sweat glands, hair follicles and sebaceous glands and concluded that pathological basis for hematohidrosis might be distinctive vasculitis. Till date there is no specific treatment. Manonukul et al, used lorazepam as anxiolytic in a case and got excellent result. Zhaoyue et al used propranolol with the hypothesis of sympathetic over activity and it was found to be effective. In a case of hematohidrosis diazepam did not work, but got excellent result with propranolol. The key to successful treatment also includes convincing the parents about the nature of the illness, it’s aggravating factors and the possible treatment of this condition.

RESULTS

All the eight cases were under mental stress. 5 cases had ear bleeding, 1 case nose bleed. Cases 7 and 8 were under intense stress and each episodes occured before examinations. Case 8 had ear, nose, oral and gastrointestinal bleed for which ENT examination, hematological evaluation, upper gastrointestinal endoscopy and laprotomy was normal. As this condition of hematohidrosis was not aware among specialist till recently, we treated with the patients with tranexamic acid and alprazolam. Patients were given counselling on life style management and reassurance. No further episodes were encountered till 3 months follow up after this clinical condition was diagnosed.

DISCUSSION

Hematohidrosis is a rare clinical condition that manifests as self-limiting episodes of spontaneous discharge of bloody secretion through intact skin or sweat gland orifices without an unknown cause. Hysteric and psychosomatic disorders are also believed to induce bleeding. Psychogenic purpura is supposed to be caused by hypersensitivity to the patient’s own blood or autoerythrocyte sensitization and is characterized by repeated crops of ecchymoses, gastrointestinal bleedings and hematuria. Another type of bleeding through skin is psychogenic stigmata, a term used to signify areas of scar, open wounds or through unbroken skin. Patients belonging to this group were found to be neurotic. Copeland reported a patient who developed bleedings from her old scars whenever she had serious anxiety. The etiopathogenesis proposed by Dr. Frederick Zugibe is that multiple blood vessels around the sweat glands arranged in net-like form. It is believed that under pressure of great stress the vessels contract. Subsequently as the anxiety passes the blood vessels dilate to the point of rupture. The blood at this point goes into the sweat glands, which pushes the blood to the surface and manifests as droplets of blood mixed with sweat. The extravasated blood has identical cell components as that of peripheral blood. The severe mental anxiety activates the sympathetic nervous system to invoke the stress-fight or flight reaction to such a degree as to cause haemorrhage of the vessels supplying the sweat glands into the ducts of sweat glands. Effect on the body is weakness and mild to moderate dehydration from the severe anxiety and both blood and sweat loss. Skin histopathological study by Zhang et al, revealed some intradermal bleeding and obstructed capillaries. No abnormality was found in sweat glands, hair follicles and sebaceous glands and concluded that pathological basis for hematohidrosis might be distinctive vasculitis. Till date there is no specific treatment. Manonukul et al, used lorazepam as anxiolytic in a case and got excellent result. Zhaoyue et al used propranolol with the hypothesis of sympathetic over activity and it was found to be effective. In a case of hematohidrosis diazepam did not work, but got excellent result with propranolol. The key to successful treatment also includes convincing the parents about the nature of the illness, it’s aggravating factors and the possible treatment of this condition.

CONCLUSION

Hematohidrosis is relatively unknown condition to the otorhinolaryngologists and the cases are occurring sporadically which goes unnoticed. This condition occurs in severe stress which may also lead to ear, nose, oral, gastrointestinal bleeds or in combination from any of the sites with a relation of severity of stress level with quantity and site of the bleeding episodes. The condition can be managed effectively if the condition is recognized after ruling out bleeding diathesis with anxiolytics and life style management counseling. If awareness is created about this clinical condition which is relatively unknown entity, it will lead to further insight on evaluation and management of this rare entity probably an exocrine disorder.
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REFERENCES
