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Review Article

Intriguing haemorrhages: a review of last seven decades (1951-2021)

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ABSTRACT

Bleeding as a cause of underlying psychological or psychiatric disorder is hardly ever thought of mainly due to unawareness and rarity. An overview will help to understand the real problem. A detailed literature survey of different types of bleeding from various parts of the body for which the reasons could not be diagnosed easily was made over last seven decades (1951-2021). These have been presented mainly in tabular form and also, in the text as briefly as possible to have a comprehensive overview only. The contents portray the nature, triggers and spectrum of settings in which haemorrhages occurred including the explanations which were available from the articles. In addition, some of my personal conjectures as possible causes have also been included. Unexplained bleeding might be psychological or psychiatric in origin. Awareness about this will enable to diagnose the case at the earliest and accordingly, the management can be started. Different types of such haemorrhage including particularly those due to idiopathic causes are potential areas of research.

Keywords: Munchausen, Factitious, Haemolacria, Haematohidrosis, Bleeding, Haemorrhage

INTRODUCTION

Psychological or psychiatric disorders cause a variety of abnormal symptoms and signs with some masquerading as underlying organic diseases which need to be ruled out first. Among all symptoms and signs, haemorrhage is the most alarming issue both to the patients and the medical caregivers - the more the bleeding, the more the distress. Normally, bleeding from any area does not occur in any individual unless there is any disease or medication including other conditions which can cause disturbances of the robust haemostatic mechanisms that prevent bleeding and/or coagulation. While many clinical features including diseases belong to psychosomatic disorders, bleeding as a manifestation of any underlying psychological or psychiatric disease is hardly ever thought of in the day to day patients' care services. In the journals, either case report or review involving one particular issue is found. This article provides a comprehensive overview of bleeding resulting from factitious, psychological, psychiatric, factitious and idiopathic causes from different parts of the body as a whole over last seven decades.

METHODS

A detailed literature survey for apparently mysterious type of haemorrhage was made from 1951 to 2021. These were analyzed. Different types of bleeding which were reported as case reports have been presented in a tabular form age-wise as per title of the concerned articles for easy understanding with minimum possible references. Those which were not covered in the tables have been included in the text as deemed relevant under different headlines. Since all routine and special laboratory investigations were done in each case to rule out any known cause, these were not mentioned.

RESULTS

Munchausen syndrome (MS)

MS is a factitious psychiatric disorder in which the individual adopts behaviours to play the role of a sick patient by feigning the symptoms or signs of a physical illness, disease or injury or faking with different biological samples in a number of deceptive ways deliberately and repeatedly in order to undergo diagnostic tests and hospitalization with medical or surgical treatment. The syndrome was named after Baron Munchausen, a eighteenth century narrator of false and exaggerated exploits. This syndrome was reported first with three cases by Richard Asher in 1951 after the patients attended with apparent acute illnesses supported by a plausible and dramatic histories which were largely made up of falsehoods.[1] MS is also known as factitious disorder imposed on self (FDIS) as per Diagnostic and Statistical Manual (DSM)-5. A few are described below.

In 1954, a 22 year old male was brought to the casualty from lying on the street as a suspected case of skull fracture with bleeding from ear and nose, past history of swelling in left hypochondrium with bleeding from ear, nose and skin, splenectomy and appendicectomy scars. While admitted, he had macroscopic haematuria with cystoscopy showing bleeding from right kidney. He was uncooperative and unruly, refused to stay in bed, wandering around the hospital usually with his face and clothes liberally smeared with blood and was discharged with a diagnosis of psychopathic malingerer. Subsequent enquiries revealed that the patient had visited 29 hospitals in British Isles, appendicectomy done in Paris, travelled widely in Finland, Ireland and on the European continent. He rarely visited the same hospital twice. Apart from Asher, this case was described by others.[2]

There are three varieties of MS – abdominal type (laparotomophilia migrans), bleeding type (haemorrhagica histrionica) and the type specializing in faints, fits, palsies etc. (neurologica diabolica).[3] The above case was an example of haemorrhagica histrionica.

Table 1: Munchausen syndrome.

Age (years)/ Gender	Brief Clinical Features	Cause/Diagnosis	Management	Reporting place
9/F	Bleeding from multiple sites – severe pain in right breast followed by bloody discharge from right nipple; parents noted similar bleeding from eyes, nose, ears, oral mucosa and umbilicus; self-limiting; no psychological stressor.	Child applying mother's liquid vermilion for faking	Refusal by parents to consult psychiatrist.[24]	NOIDA[a]
10/F	Sudden bloody discharge from right eye in the evening when alone in a dark room for 2 weeks; follow up every 3 days with instruction to come whenever bleeding occurred; after 4 days, "emotionally distraught" father brought the child to inform blood streaking linearly on her right cheek on the previous evening.	Biting of buccal mucosa on right side, taking out the blood with her fingers and dabbing on her cheek to gain attention	Not available.[25]	Allahabad[a]
12/F	History of haematemesis and epigastric pain -no cause found by gastroenterologist; consulted another hospital for similar complaints after 4 weeks – no cause; developed persistent productive cough with moderate amount of blood; referred back to previous hospital for respirology service; two episodes of epistaxis coinciding with first episode of haemoptysis; cheerful girl and model student; ulcerated lesion in left nostril but no active bleeding; when left alone with parents at the examination room, expectorated about 30 ml of bloody fluid; readmitted; numerous fresh-appearing lesions- deep on the innermost aspect of lower lip mucosa consistent with biting ; 1st evening-episodes of haemoptysis; 2nd evening– patient forcibly driving her finger into her nostril and then placing the blood from her finger into her mouth while holding the calling bell as witnessed by a nurse.	Bullying by children at school, significant conflict with mother and upsetting with nurse caused self-inflicted injuries	Referred to Psychiatry department.[26]	Alberta (Canada)

Continued.

Age (years)/ Gender	Brief Clinical Features	Cause/Diagnosis	Management	Reporting place
16/F	Blood coming from both eyes admixed with tears for 1 day; streaks of blood from both fornices; hospitalized; bleeding occurred when alone; moved to a bed near nurses' station; when bleeding was noted, medical team rushed to the spot – blood streaking from both eyes and cheeks in a linear pattern.	Puncturing left side finger tips using hair clip with right hand for faking	Discharged next day; did not attend follow up visits.[27]	Amnan (Jordon)
19/F	Complaints of 3-4 episodes of sudden, bright red and severe bleeding without any symptoms from right ear when she was alone in her room for the last one week.	Self-inflicted trauma and emotional stress from some family issues (as disclosed by her mother)	Counselling; advise to come with photographs whenever bleeding. No recurrence.[2]	Karachi (Pakistan)
27/F	Presented with haematuria; treated for the same by many; laparoscopic right nephrectomy for arteriovenous malformation; post surgery, macroscopic haematuria continued but the cause remaining unclear; received more than 200 units packed cell for severe anemia and frequent septicaemia.; no improvement of anemia; during transfusion, haematuria started.	Injecting some of the transfusion blood into her own bladder using hidden dirty syringes	Automatically discharged herself (mother informed at that time that she held nursing license).[29]	Yufu (Japan)
28/F	Bleeding from a tracheostomy site with front gown soaked with blood when discharge was being planned; after 3 days, severe episode of bleeding, the force of which was sufficient to propel to ceiling and was found so; showed several blood-stained gauzes; patient moved to a place for observation by nurses.	Drawing blood from a vein in her arm and then spattering it around her neck and ceiling	Outpatient management by psychiatrist.[30]	Columbia (US)
30/F	One day history of severe cramps, abdominal pain and 5 episodes of bloody diarrhoea; admitted; on the 3rd day - vaginal bleeding, persistence of bloody diarrhoea and severe abdominal pain unrelieved by antibiotics; persistent symptoms; false history of occupation and ulcerative colitis.	Injecting blood into rectum, vagina and pouring into the commode admixing with stools after drawing the same from intravenous or peripherally inserted central catheter line and then storing in the syringes hidden under her bed	Refused consultation with psychiatrist.[31]	Milwaukee (US)
46/M	History of fall at workplace 4 years back with loss of consciousness and bleeding from right ear - managed symptomatically; multiple hospital visits for recurrent bleeding from right ear and hearing loss; extensive scratch marks and blood clots in posteroinferior quadrant of external auditory canal in right ear; bony canal and middle ear inspected; cortical mastoidectomy done; bleeding complaint persisting; whenever discharge was advised, reported jerky movements of all four limbs, staring at objects and refusing to take medicine; extensive review over 4 years.	Separated from wife, financial problems in educating three children	Monthly follow up at Psychiatry department; no recurrence.[32]	Thimphu (Bhutan)

[a] India

In addition to those presented in Table 1, MS masqueraded as a sudden and intermittent spontaneous bleeding of about 1 to 2 ml blood from forehead, face, cheek, eyes and scalp in a 7 year old girl first without any relevant history and subsequent relevant investigations for the same failed to reveal any cause followed by occurrence of the same type of problems from 4 other children from the same locality and socio-cultural background. The most common site was scalp. Microscopic examination of the blood from the affected areas revealed plenty of mucous strands and epithelial cells. Later, 2 fresh cases came to the hospital from a different locality where the index case visited. Covert video surveillance and interviewing revealed biting of buccal mucosa and then applying the spitted blood on the forehead briskly and smartly. It could not be ascertained from their uncooperative mothers' reaction as to whether they were aware of the problem or had been assisting them. While the index case was proved to be MS, others might be Munchausen syndrome by proxy as explained below.[4]

Munchausen syndrome by proxy (MSBP)

MSBP, also known as factitious disorder imposed on another (FDIA) as per DSM-5 is a parenting psychological disorder in which a parent, almost invariably the mother causes the following on her child either alone or in combination - harm, induction or fabrication of an illness, falsification of medical history, tampering with the medical specimen, deception with fictitious or exaggerated information etc. in order to create a situation that requires or seems to require medical attention. The term, MSBP was coined by Roy Meadow in 1977.[5] MSBP is also known as Meadow's syndrome, Polle Syndrome, fabricated illness by proxy and factitious disorder by proxy. Patients with MSBP undergo innumerable unnecessary, even harmful procedures including prolonged hospital stay and multiple drug administrations with undesired consequences. Some cases have been described in Table 2. 19 such cases including 2 deaths, aged 4 months to 7 years (10 boys and 9 girls) from 17 families were reported in which episodes of bleeding from many areas, neurological abnormality, rashes, fevers and abnormal urine were cunningly simulated by the mothers on their children ranging from 1½ months to 4 years with one examined by 28 doctors. Self-pricking, stirring her vaginal tampon during menstruation into the child's urine specimen or substituting her own pathological urine for the child's urine, mixing stool with child's vomit, rubbing thermometers or immersing those in hot fluid, diluting or adding chemicals to blood specimens, rubbing skin gently and repetitively with finger nail or sharp objects, applying caustic solutions, adding paint, cocoa, phenolphthalein etc., giving sedatives and tranquillizers with high doses and applying pressure on carotid sinus were the various ways adopted by the 16 culprit mothers which included 8 nurses.[6] Surprisingly, the mother spat and placed blood in her child's ear and video recorded the result to gain medical attention.[7] In another case, the mother simulated symptoms including a 2 month history of bloody vomiting, bloody stool and nose-mouth-ear-eye bleeding by contaminating blood to her 8 months daughter. She complained of multiple vaginal bleedings but no bleeding when hospitalized and the source of blood in both of them could not be determined.[8] A review of 117 cases revealed bleeding was one common presentation among others with abstruse mothers being the perpetrators in all cases with 100% short term morbidity, 14% failure to thrive, 8% long term morbidity and 9% mortality rate.[9]

Table 2: Munchuasen syndrome by proxy.

Age (years)/ Gender	Brief Clinical Features	Cause/Diagnosis	Management	Reporting place
8 mo / F	Admitted for blood vomiting for 2 months; reluctance by mother while taking history; endoscopy revealed linear hemorrhagic lesions at tonsils and pharynx.	Doctor witnessed - mother was injuring the child with her finger	Warning to mother about security footage; resolved.[33]	Ankara[b]
8 mo/F	History of intermittent haemorrhage in ear, nose and eyes for 2 months and in stool for a few days, also in vomitus; coagulums found on the outer and front part of the nose; repeated symptoms; no clue despite highly specialized investigations.	Mother resorted to the method for problems with the mother-in-law	Follow-up of mother at Psychiatry department.[34]	Bolu[b]
1y1mo/ M	Admitted for recurrent haemoptysis for 4 months; occurrence when with mother alone; while hospitalized, fresh bleedings and blood-stained clothes.	Security footage – mother sneakily obtained blood from other patients for deceiving	Warning to mother; resolved.[33]	Ankara[b]

Continued.

Age (years)/ Gender	Brief Clinical Features	Cause/Diagnosis	Management	Reporting place
2/M	History of bleeding for 2 months with multiple consultations; colonoscopy - small rectal ulcer 2 cms away from the opening with a reddish halo and excoriation in the perianal region; anxious mother.	Inflicting injury to rectal mucosa by mother using finger tips as advised by her friend to have an opportunity to avoid staying at home due to conflicts with husband and family.	Persuasion for psychiatric treatment failed; demanded immediate discharge for fear of severe punishment.[35]	Vellore[a]
9/M	Complaint of fit-like episodes for last 3 months and haematemesis for 1 year with multiple admissions; fit-like episodes despite on antiepileptics; father and paternal uncle used to stay with the child, anxious, demanded more investigations; 2-3 fit-like episodes but not seizures and repeated haematemesis while in ward; father collected sample in bottle to show doctor; no RBC but salivary secretions mixed with some reddish-brown chemical.	Fabrication of haematemesis by his father and uncle by giving him betadine just before vomiting for dysfunctional family life.	Admitted in psychiatry; no haematemesis for 1 week; offenders were confronted but denied; social services decided to keep the child under mother.[36]	Rohtak [a]

[b] Turkey, [a] India

Table 3: Haemolacria

Age (years)/ Gender	Brief Clinical Features	Cause / Diagnosis	Management	Reporting place
6 months/F	Bloody tears from both eyes for three days for the first time; flu-like illness 1 week before; yellowish discharge from birth; clinical examinations and all relevant investigations – normal .	Undetermined source	Fusidic acid eye drop-1%; resolved within 3-4 days; no recurrence.[37]	Dhahran (Saudi Arabia)
10/F	Spontaneous, transient, recurrent, 5-6 times within same day - painless and self limiting bleeding from eyes for last 3 months; pampered; after admission - crying spells, irritable, social withdrawal, difficult rapport establishment, silent; divulged - bleeding when not happy.	Childhood depression	Sertraline –25 mg h.s., then lorazepam i.m., clonazepam – 0.25mg b.i.d., propranolol–10 mg t.i.d.; after 2 days bleeding stopped.[38]	Chennai[a]
11/F	Bilateral spontaneous blood tinged tears occurring 2-3 times a day lasting for 2-3 minutes and not associated with any stress or cry for 2 weeks; past history of epistaxis; direct observation for 2 days revealed the above also.	Idiopathic	Family counselling; under follow up.[39]	New Delhi[a]

Continued.

Age (years)/ Gender	Brief Clinical Features	Cause / Diagnosis	Management	Reporting place
15/F	1st episode -bloody tears when under stress of house examination; 2nd episode- trickling over cheeks while concentrating on books accompanied by headache and giddiness; 3rd episode-bilateral haemolacria trickling over both cheeks observed by the authors while admitted total -11 episodes occurred.	Hysterical traits	Reassurance, vit C, K & injectable B complex – ineffective.[40]	Rohtak[a]
16/F	Intermittent, irregular, bilateral bloody tears for 3 months.	Idiopathic	Regular follow up only.[41]	Samsun (Turkey)
22/M	Two episodes of unilateral spontaneous bloody tears; normal functionality of von Willebrand factor with a glycoprotein binding assay, ristocetin cofactor activity and ristocetin-induced platelet agglutination assays plus non contributory in-depth psychosocial analysis.	Idiopathic	NA.[42]	Port Blair[a]
25/F	Crying red tears with similar episode plus mild epistaxis one month back coinciding both with menstruation.	Ocular vicarious menstruation	Oestrogen-progesterone oral contraceptive pills; no recurrence in 3 months.[23]	Chandigarh[a]

[a] India, NA – Not available

Table 4: Haemolacria and haematohidrosis.

Age (years)/ Gender	Brief Clinical Features	Cause / Diagnosis	Management	Reporting place
13/F	Bleeding from the face, eyes, shoulders and forearm for 1 hour with a history of such attacks preceded by anxiety for 2½ months.	NA	Anxiolytic and propranolol– symptom free.[43]	Kolkata[a]
15/F	Profuse bleeding from the ears, nose and eyes for last 2 weeks following a head injury; relevant investigations did not show any sign of trauma or source of bleeding.	NA	Anxiolytic and propranolol – symptoms reduced, not stopped.[43]	Kolkata[a]

[a] India , NA–Not available

Table 5: Haemolacria with another bleeding site.

Age (years)/Gender	Brief Clinical Features	Cause / Diagnosis	Management	Reporting place
13/F	Recurrent episodes of spontaneous bleeding from eyes and ears lasting for 4-5 minutes over 2 years.	Essential idiopathic	Family counselling, reassurance and follow up.[44]	New Delhi (India)

Continued.

Age (years)/Gender	Brief Clinical Features	Cause / Diagnosis	Management	Reporting place
16 /F	Haemolacria with epistaxis accompanied by headache and lower limb cramps with 3 years of acquired immunodeficiency syndrome; during hospitalization, two episodes of the same as above.	Idiopathic	Antiretrovirals.[45]	La Havana (Cuba)
17/F	Spontaneous bleeding from both eyes and gum while crying for 15 days with abnormal twitching movement of whole body and shortness of breathing.	Idiopathic with associated psudoseizure and psychogenic hyperventilation	Amitryptiline – 10 mg h.s. and counselling; no recurrence in 6 months.[46]	Jeshore (Bangladesh)

Table 6: Haemolacria in combination with bleeding from multiple areas.

Age (years)/ Gender	Brief Clinical Features	Cause / Diagnosis	Management	Reporting place
14/F	Episodes of burning sensation all over the body followed by bizarre behaviour and unconsciousness; episodic bleeding from mouth, eyes and ears plus self-mutilating behaviour particularly chewing of lower lip and stiffness in the lower limbs for 2 months after a family stress; at her 6 years of age, she had episodic respiratory distress with increased frequency not responding to treatment and subsequently, with a variety of new features.	Mixed dissociative disorder	Low dose antidepressant, explanation to the patient and the family. Symptom free within 1 month and remained so for 1 year; recurrence on reexposure to family stressor; anxiolytics and interpersonal psychotherapy.[47]	Sylhet[c]
16/M	Multiple episodes of bleeding from both eyes, mouth and urethra, chest pain, severe convulsion and inability to talk with a history of episodes of different type of features for 2 years– pricking sensation on the left side of the body followed by breathlessness, multiple episodes of abnormalities characterized by unresponsiveness, convulsions, loss of power in the limbs, inability to hear and see the bleeding from nose and eyes; cutting hands to bleed.	-do-	Minor improvement with antidepressant & anxiolytics; later additionally individual therapy addressed conflicts, behavioural principle and family therapy - no recurrence for 1 year.[47]	Sylhet[c]
17/F	Episodes of bilateral haemolacria, abnormal abdominal movement followed by blood vomiting after food and paroxysms of abdominal pain for 2½ years with a history of episodes of dizziness, headache and vomiting for 1½ months 4 years back, visual and auditory hallucinations, loss of appetite and vomiting after meal, increasingly demanding and aggressive behaviour and then episodes of convulsions, episodic loss of consciousness and inability to remember anything after	Mixed dissociative disorder and migraine	Anxiolytics and low dose antidepressants for a short period; then interpersonal psychotherapy with explanation of her problems, discussed targeting the patient and family; no sequel up to 2 years.[47]	Sylhet[c]

Continued.

Age (years)/ Gender	Brief Clinical Features	Cause / Diagnosis	Management	Reporting place
	recovery for about 4½ years plus infrequent migraine attacks on exposure to sunlight.			

[c] Bangladesh

Table 7: Haematohidrosis.

Age (years)/Gender	Brief Clinical Features	Cause / Diagnosis	Management	Reporting place
8/M	Spontaneous recovery of painless oozing of bloody secretions from nail beds from hands and feet.	Fear of upcoming examinations, watching fiction movies and when parents not satisfying demands	Parents provided with information about benign and self-limited disorder; no episode in 2 years.[48]	Tehran (Iran)
9/F	Several episodes of spontaneous, unpredictable, intermittent oozing of blood from ear lobules, nose, eyes with no specific pattern and ceasing also spontaneously after 5 minutes.	NA	Propranolol -10 mg twice daily; significant improvement with no episode in 6 months.[48]	Tehran (Iran)
9/F	Recurrent episodes of self-limited spontaneous bleeding from skin, scalp, ear, mouth and eyes - twice weekly and mostly, in the evening lasting for 1 to 2 minutes without any precipitating factor; no episode on holidays.	Bullying at school by her classmates	Propranolol – 10 mg twice daily; resolved.[49]	Riyadh (Saudi Arabia)
10/F	Oozing of blood from intact skin of scalp once or twice a day lasting for about 3-5 minutes for 1 week with starting of spontaneous bleeding due to punishment by the teacher to stand outside the class for misconduct after about 30 minutes; fearful of father; scolding by both parents for being slightly less scholastic underperformer than her younger sibling; nocturnal enuresis for 2 years about once or twice a week, almost daily during examination time and when scolded preceded by 5 years of continence.	Mixed anxiety and depressive disorder	Imipramine, clonazepam, reassurance and relaxation exercises, psychoeducation of parents; bleeding stopped after 4 months.[22]	Chennai[a]
11/F	Spontaneous, recurrent and painless bleeding from forehead, eyes, ears, nails, arm, umbilical area, back, vagina and gastrointestinal tract - most of these preceded by abdominal pain, vomiting or headache but sometimes occurring spontaneously or during sleep with subsidence on its own after 10-15 minutes for several months; during admission - 2 episodes of moist, viscous, bubbly blood stained sweaty discharge from limited area of forearm but no oozing after wiping.	Extreme physical or emotional stress	Counselling; propranolol -10 mg; marked improvement only but not complete recovery in 6 months.[48]	Tehran (Iran)

Continued.

Age (years)/Gender	Brief Clinical Features	Cause / Diagnosis	Management	Reporting place
12/F	Several episodes of spontaneous, intermittent unpredictable painless bleeding from the left side of the face mostly around orbital regions, left eye and tear ducts unassociated with mood, activity or sleep for 2 weeks; two episodes were observed by the physician while admitted – starting like a tear drop confluent spots of mild watery secretions over the left side of the face and streak-like drops starting from the corner of the eye and along the cheek followed by bright red coloured secretion lasting for 10-20 minutes but patient sat showing no distress.	Idiopathic	NA.[50]	London (UK) and Jabriya (Kuwait)
18 /F	Multiple episodes of spontaneous, self-limited mucocutaneous bleeding from nose, lacrimal ducts, forehead, hands, nails and navel which were immediately proceeded by intense headache and abdominal pain for 6 months; more than 30 bleeding episodes –witnessed while hospitalized.	Stress	Propranolol[51]	Alicante (Spain)
21/F	Self-limited episode of bleeding from palms and face lasting for 1 to 5 minutes for 3 years; occurrence while working and sleeping; more so during perceived emotional stress.	Depression, anxiety, major depressive and panic disorder	Paroxetine and clonazepam; bleeding continued; propranolol- 20 mg/day; marked reduction only.[52]	Florence (Italy)
72/M	Haematohidrosis involving the anterior part of the lower abdomen for 2 months.	Depressive disorder	Counseling only; no recurrence till 1½ years.[18]	Mumbai[a]

[a] India, NA – Not available

Table 8: Miscellaneous (Bleeding from other sites).

Age (years)/Gender	Brief Clinical Features	Cause / Diagnosis	Management	Reporting place
20 months/F	Brought by mother for bilateral breast development; Tanner stage 2 but no pubic hair; relevant hormonal investigations - pubertal level; diagnosed premature thelarche; monitoring; 1 month later, brought for vaginal bleeding; repeat hormonal level- prepubertal, clinical and radiological examinations- nonindicative; during next 6 months, recurrent vaginal bleeding every month but regressed breast development with normal growth velocity; bleeding - concurrent with her mother's menstrual period; referral to a psychiatrist; mother	Factitious disorder	Psychiatric disorder of mother– undiagnosed.[53]	Ankara and Samsun (Turkey)

Continued.

Age (years)/Gender	Brief Clinical Features	Cause / Diagnosis	Management	Reporting place
	confessed applying her vaginal flow to daughter's underwear.			
10/M	Repeated bouts of haemoptysis, haematemesis, epistaxis, haematochezia and haematuria for 3 months preceded by haemoptysis while in school; then repeated episodes of oozing of blood from navel, eyes, ear lobules and nose while at home or school; usually with haemolacria when there is upcoming examination, parents not satisfying demands etc.; pampered, adamant and defiant.	Haematohidrosis with oppositional defiant disorder	Lorazepam–1mg h.s. and propranolol–10 mg b.i.d for 3 weeks, then only propranolol; graded behavioural intervention and relaxation exercises for child, psychoeducation of parents for positive reinforcement; bleeding stopped with normalization of symptoms.[54]	Vijayawada[a]
11/F	History of finding of blood stains on her sleeves by the mother; lesions on the forearm-linear, horizontal with tailing towards right, on the forehead-vertical tailing down; patient claimed spontaneous appearance of those over last 7 days but was relaxed and with unusual indifference.	Dermatitis artefacta	Fluoxetine-10mg, psychotherpy, family therapy; no recurrence for 1 month.[55]	Kolkata[a]
15/F	Recurrent bilateral upper extremity and torso bruising associated with general malaise, headache and joint pain; correlation with mood changes, low self-esteem, academic decline, poor interpersonal relationship, heightened feelings of stress with ultimate initiation of the lesion with ongoing parental divorce proceedings.	Major depressive disorder with psychogenic purpura	Escitalopram–10 mg and intensive supportive therapy; resolution of symptoms and improvement of mood changes after 3 months.[56]	Michigan (Canada)
15/F	Episodic bleeding from nose and both ears including decreased sleep - 8 months; episodes of fits -10 months and headache-2 years; after sustaining an accident 2 years back, headache off and on; more demanding, easily becomes angry and impulsive; headache - after crying when hurt, read for a long time and exposed to heat followed by dizziness, dimness and blurring of vision, hearing loss and then unconsciousness; bleeding from nose and ears	Conversion disorder and migraine with anxiety features	No treatment mentioned but symptom free for 1 year.[47]	Sylhet(Bangladesh

Continued.

Age (years)/Gender	Brief Clinical Features	Cause / Diagnosis	Management	Reporting place
	with recovery after a brief period.			
15/F	Recurrent episodes of fissuring, bleeding and haemorrhagic crusting of the lower lip for last 7 months; flare-up ranging from minor fissuring and erosion to frank bleeding while encountering relationship issues with parents and siblings; punch biopsy - chronic inflammation; overprotection by the parents, borderline personality traits, impulsivity and poor frustration tolerance.	Factitious chelitis	Supportive psychotherapy, parental counselling, environment manipulation, fluoxetine, topical emollient and occlusion; regression.[57]	Pondicherry[a]
18/F	Abrupt onset of bleeding from the nose; nonresponse to medication; referred to Psychiatry; Gynaecology- multiple superficial cut marks- 1cm x 5 cm over thigh and upper leg on medial aspects, few recent; mother observed – she was self-injuring with sharp objects in unexposed areas and mixing it with her nasal secretions.	Significant ongoing stress due to parental conflict; seeking parental attention	Fluoxetine and cognitive behavioural therapy; fluoxetine stopped after tapering for 9 months.[58]	Kerala[a]
25/F	Complaining of recurrent gastrointestinal bleeding starting with rectal blood loss 4 years back with a history of admission in another hospital for rectal blood loss with colonoscopy twice; endoscopic examinations- negative; Hb- 8.7g/dl; during hospitalization, more rectal bleeding with more frequency; colicky abdominal pain preceded rectal bleeding; colonoscopy twice- red blood was seen in ascending, transverse and descending colon but no lesion; pertechnate abdominal scintigraphy – no ectopic gastric mucosa; transfusion of 16 units of blood in 3 weeks; scintigraphy with labelled RBC – no clue; after half an hour of leaving the patient alone, colicky abdominal pain and passing red blood per rectum; images taken during this time showed bleeding in descending colon but the passed blood was not radioactive; physical examination revealed injection marks in the left lower	Factitious bleeding due to self-inflicted illness and injuries using several needles and syringes together containing 1 pint of blood (found in cupboard); self-administration of enema of blood drawn previously from infusion bottle; during scintigraphic examination, managed to draw blood from her arm unnoticed and injected intraabdominally.	NA.[59]	Leiden (Netherlands)

Age (years)/Gender	Brief Clinical Features	Cause / Diagnosis	Management	Reporting place
	abdominal wall; formerly a student of nursing.			
84/M	Complaint of pins and needles affecting the left arm, collapse with severe left sided weakness after hearing the death news of his hospitalized wife; hypertension on methyldopa; GCS–E3V2M5, Cheyne-Stokes breathing, BP–198/102 mm Hg, P–62/m, 2 mm reactive pupil, dysconjugate gaze paresis, complete left hemiparesis; CT scan revealed right thalamic/mid brain haemorrhage.	Primary intracerebral haemorrhage due to sudden emotional upset	Supportive treatment;death-5 days after admission.[21]	Scotland (UK)

[a]India

Haemolacria

Haemolacria is bloody tears ranging from red-tinge to blood. This is also known as dacryohaemorrhoea, haematodacryorrhoea and sanguineous lacrimation. It can be unilateral or bilateral. The details of individual case reports are in Table 3-6. In addition, there was no cause in 4 cases with recurrent unilateral episode (6-14 years) but resolved spontaneously.[10] Also, out of 27 cases of spontaneous haemolacria over 3½ years (2015-2018), only 5 patients <18 years old had idiopathic causes.[11] Examination of occult blood in tears in 125 healthy subjects revealed startling observations of occult haemolacria in 18% of the fertile women (n=64) most often in the menstrual phase, 7% pregnant women (n=30), 8% in male (n=24) and none at the menopause (n=7).[12]

Haematohidrosis

It is also known as haematidrosis which means sweating/oozing of blood from intact skin. Nine case reports are there in Table 7. A review of haematidrosis (1996-2016) with 25 cases revealed 84% were females with 62% from Asia, mainly India and bleeding sites were face (96%) including forehead (40%) apart from other areas either alone or in combination, the causes being mainly stress together with epilepsy and platelet dysfunction in only two cases.[13] Sibling bleeding as haematohidrosis (9 year old boy and his six month old sister) was reported for the first time with a history of bleeding episodes from ears, eyes, scalp and other sites which showed positive response to propranolol with decreased frequency and severity of bleeding.[14]

Miscellaneous (Bleeding from other sites)

Nine case reports from various sites due to different causes have been indicated in Table 8. Among many such reports including reviews, a few are described below. A 16 year old female with complaints of bleeding from multiple and unusual sites including haemoptysis, haematuria, haemolacria and bloody nipple discharge only witnessed by the mother was diagnosed to be MSBP by a multidisciplinary team.[15] History of spontaneous haemorrhages from eyes, scalp, hands, feet, ears, nose, scalp and in urine occurring up to 20 times daily particularly dripping from eyes down her face in a 13 year girl while the patient was calm and indifferent drew international media attention including filming of a National Geographic Channel documentary in 2009 ("Twinkle bleeding"). However, nobody has seen the initiation except the mother, bleeding episodes decreased during irregular and heavy menstrual period which lasted for 10 days and both the patient and her mother refused to stay as inpatient including psychiatric consultation. Hence, no diagnosis could be made but it was likely to be a case of factitious disorder.[16]

DISCUSSION

Only some patients described under different tables and in the text briefly portray the nature, triggers and spectrum of settings in which bleeding occurred to make this article short.

Haemorrhage from anywhere particularly from unusual and multiple sites and/or mixed with normal body fluids is usually exaggerated by the patients, more so by the relatives precluding to ascertain the nature and amount of bleeding. The situations became, at times, flabbergasting to the treating doctors including expenditure on resources particularly for factitious disorders. Bleeding as a cause of mental disorder is neither generally taught nor usually discussed in academic forums except stress ulcers or uterine bleeding. Hence, this is hardly ever considered. Minimization of symptoms or cure by appropriate drugs, mostly related to antidepressants, anxiolytics and beta blockers validates provisional/ final diagnosis or points to the possible cause in individual cases. Idiopathic cases and those patients who responded partially or not at all are due to hitherto incomplete understanding. Gene-environment interaction might be the cause in the predisposed persons.

The exact cause of MS is not known but could be due to parental neglect, emotional trauma or illness in childhood with unresolved issues, personality disorder etc. The same may also be applicable for MSBP but seemingly, more complex because normally, even in animals, the mother tries to protect their babies. Hence, MSBP is possible only by unsuspecting mother with covert psychiatric problem or seriously bewildering psychological imbalance resulting from overpowering natural emotion of the mother to inflict injuries unnecessarily and repeatedly. After one year of joining our hospital 29 years back, I heard about an employee aged about 40 years who used to mix slight beetroot extracts to urine to fake haematuria to avoid transfer to another steel plant and in another hospital, one 55 year aged female who used to fake the same by self-injuring gum near lower molar teeth using a safety pin kept hidden inside her blouse to teach her daughter-in-law for misbehaviour and son's inability to control his wife. Both perhaps suffered from MS. Some features of MS would be absent in children mainly due to their dependence on parents.

Psychogenic purpura, also known as Gardner–Diamond or autoerythrocyte sensitization syndrome is a psychological or dermatologic syndrome involving spontaneous and painful ecchymotic purpuric lesions that typically appear after a period of stress or minor trauma mostly on the extremities including spontaneous appearance of recurrent bruising, multi-organ bleeding like menorrhagia, haematuria, epistaxis, haemolacria, and gastrointestinal bleeding and usually in young adult women with emotional disorder. Although the above syndrome was named in 1955, the first such report dates back to 1927.[17]

Normally, stress induced activation of sympathomedullary system results in net hypercoagulability as a protective mechanism but in haematohidrosis, it is due to dermal capillary blood vessels rupture under extreme emotional or physical stress due to exacerbated sympathetic activation leading to periglandular vessel constriction and subsequent expansion including possibility of dermal stromal weakness which allows the blood vessels to act like wax and wane of a balloon allowing the passage of blood contents into ducts.[18,19] Observation of stress induced bleeding episode due to qualitative von Willebrand factor deficiency (Type 2) indicates psychobiological link between the two.[20] Sudden changes in blood flow/pressure are accompanied by rapid compensatory mechanisms but this does not happen with ageing. In cerebral haemorrhage, perhaps due to unique susceptibility secondary to genetically determined variability in vascular responsiveness plus acquired pathology, some vessels affected by fibrinoid necrosis occlude or rupture while some held without sequelae.[21] Bleeding in mixed anxiety and depressive disorder occurs due to underlying intense fear secondary to psychosocial stressor.[22]

The pathology of vicarious menstruation could be due to increased permeability and congestion resulting from cyclical hormonal changes even in the absence of endometrial tissue at extrauterine sites including cyclical variation of conjuctival epithelium or nervous system instability.[23] Although cyclical bleeding from nasolacrimal endometriosis is associated with haemolacria, other forms of extragenital endometriosis may occur non-cyclically. Occult haemolacria in females during fertile period seems to indicate involvement sex steroid hormones and appears to be the harbinger of vicarious menstruation if the degree of underlying cause for the same attains a threshold level.

It is also perplexing as to why unilateral haemolacria is observed in some cases for the same reason. This is presumably due to differential somatization. Similarly, the exact cause for site-specific haematohidrosis and bleeding from other areas might be resulting from individualized somatization due to underlying complex neurohumeral mechanism with the same stress. Hence, these areas need to be explored in detail.

These afore-mentioned cases are, indeed, rare but with female preponderance particularly the young. The reasons for occurrence particularly in young females might be due to temporary neurohumeral disturbances with possible genetic predisposition consequent on lower psychological threshold to stress in the setting of preexisting societal gender based various restrictions including different ways of nurturing from the birth.

Although the review has been made for last seven decades only with increasing number of reporting with the passage of time from many countries, historically, such cases were reported very rarely particularly haematohidrosis from 3rd century B.C.; in Jesus Christ before crucifixion and a soldier before going to battle by Leonardo da Vinci; haemolacria was described in the scientific medical book, Aetius of Amida, in the 6th century and then in the 16th century, there was a nun who instead of menstruating had auricular and ocular haemorrhage every month; in 1581, Dondonaeus observed that the nature attempted to rid itself of the excess blood through the eyes in a 16 year aged girl who had not menstruated and in 1755, a 15 month aged boy shed bloody tears on several occasions. The reasons for increasing reports could be due to more awareness and expression of mental problems in genetically predisposed persons in the changing socio-cultural scenario.

The role of laboratory for detecting blood mainly centers on microscopy of the specimen for RBC with or without presence of others, tests related to haemoglobin including porphyrin, checking the blood group whether matching with the patient or mother, even if matched grouping-genetic analysis to reconfirm its origin from either of the two in doubtful cases including in rare cases, of nonhuman origin.

CONCLUSION

This article is mainly for awareness. Patients of factitious disorders are intelligent with cunning ways of deceptions. Diagnosis of factitious disorders are made by labour-

intensive and time-consuming efforts, expensive procedures, careful observations keeping all possibilities in mind, finding some hidden sites and examination of the whole body. The bleeding in psychiatric disorders and idiopathic cases indicates hitherto poorly understood/unknown mechanisms but these must be associated with disturbed vascular haemodynamics, alteration of vascular permeability, abnormal control of haemostatis either through complex neural control on vessels or more probably, altered regulation of gene expressions at different levels related to production of clotting factors, platelet aggregation and maintenance of capillary endothelium. Hence, well-coordinated multidisciplinary researches are expected to unravel the mystery. In view of the above, it appears prudent to think of psychological/psychiatric disorders in doubtful cases. Anticipating progressive number of such cases in the changing socioeconomic and cultural scenario, awareness will make the diagnosis easier than before. To conclude, this is a comprehensive overview only with research potential in any of the above haemorrhages.

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REFERENCES

1. Asher R. Munchausen's syndrome. The Lancet. 1951;1:339-41.
2. Gown RA, Kauffmann EA. Munchausen Syndrome. BMJ. 1955;1068.
3. Asher R. Munchausen Syndrome. BMJ. 1955;1271.
4. Sridharan S, Shukla D, Mehta R, Oswal R. Munchausen syndrome masquerading as bleeding disorder in a group of pediatric patients. Indian J psycholo Med. 2011;33:86-8.
5. Meadow R. Munchausen by proxy. The hinterland of child abuse. Lancet. 1977;2:343-45.
6. Meadow R. Munchausen syndrome by proxy. Archives of disease in childhood. 1982;57:92-8.
7. Bennett AMD, Bennet SMV, Prinsley PR, Wickstead. Spitting in the ear: a falsified disease using video evidence. J Laryngol Otol. 2005;119:926-7.
8. Ozdemir DF, Yalcm SS, Akgui S, Evinc SG, Karhan A, Karadag F et al. Munchausen by proxy syndrome: a case series study from Turkey. J Fam Viol. 2015;30:661-71.
9. Rosenberg DA. Web of deceit: A literature review of Munchausen syndrome by proxy Child Abuse Neglect. 1987;11:547-63.
10. Ho VH, Wilson MW, Linder JS, Fleming JC, Haik BG. Bloody tears of unknown cause: case series and review of the literature. Ophthalmic Plast Reconstr Surg. 2004:20(6):442-47.
11. Bai F, Zhou XB,Wang P, Wang LH, Wang F, Tao H. Retrospective investigation of spontaneous bloody tears: a report of 27 cases, Zhonghua Yan Ke Za Zhi. 2020; 56(1):53-8.
12. Ottovay E, Norm M. Occult haemolaria in females. Acta Ophthalmol. 1991;69;44-6.
13. Kluger N. Hematidrosis (bloody sweat): a review of the recent literature (1996-2016). Acta Dermatovenerol APA. 2018;27:85-90.
14. Hoover A. Fustion N, Sparks AO, Rokes C. Sweating blood: a case series of 2 siblings with hematohidrosis, J Pediatr Hematol Oncol. 2021;43:70-2.
15. Tufekci O, Gozmen S, Yilmaz S, Hilkay Karapnar T, Cetin B, Burak Dursum O. A case with unexplained bleeding from multiple sites: Munchausen syndrome by proxy. Pediatr Hematol Oncol. 2011;28:439-43.
16. Benzer SK, Buchanan GR. Bleeding from the eyes and through intact skin: physiologic, structural, spiritual or faked. Am J Haematol. 2013;88:713-16.
17. Dick MK, Klug MH, Gunmadi PP, King LK, Huerter CJ. Gardner- Diamond syndrome: A psychodermatological condition in the setting of immunodeficiency. J Clin Aesthet Dermatol. 2019;12:44-6.
18. Jerajani HR, Jaju B, Phiske MM, Lade N. Hematohidrosis–a rare clinical phenomenon. Indian J Dermatol. 2009;54:290-92.
19. Kanel RV. Acute mental stress and hemostasis:when physiology becomes vascular harm. Thromb Res. 2015;135:S52-5.
20. Subramanian K, Pravallika M,Menon V. Evidence for stress induced bleeding in a patient with von Willebrand factor deficiency. Indian J Psychol Med. 2018;40:292-95.
21. Lammie GA, Lindley R, Keir S, Wiggam MI. Stress-related primary intracerebral haemorrhage. Stroke. 2000;31:1426-28.
22. Jayaraman AR, Kannan P, Jayanthini V. An interesting case report of hematohidrosis. Indian J Psychol Med. 2017;39:83-5.
23. Ghosh S., Tale S, Hansda N, Bhalla A. Rare case of red tears: ocular vicarious menstruation. BMJ Case Rep. 2021;14:e237294.
24. Pandey M, Sawhney A. Factitious bleeding disorder in a child: an unusual presentation of Munchausen syndrome. Indian Paediatrics. 2014;51:1019-20.
25. Chaudhary A, Agasti M. Hemolacria: a rare presentation of Munchausen syndrome. International J Ocular Oncol Oculoplast. 2016;2:211-13.
26. Bjornson CL, Kirk VG. Munchausen's syndrome presenting as hemoptysis in a 12 year old girl. Can Respir J. 2001;8:439-42.
27. Karadsheh MF. Bloody tears: A rare presentation of Munchausen syndrome case report and review. J Family Med Prim Care. 2015;4:132-34.
28. Khalil A, Abbas SA, Qureshi T, Ali S, Ayub B. A case report of Munchausen syndrome presenting with

bleeding from ear: a rare factitious disorder. J Pak Med Assoc. 2020;70:168-70.
29. Ando T, Nomura T, Sejiyama S,Shin T, Mori K, Sumino Y. Munchausen syndrome in the act of creating and enacting microscopic hematuria. Uro Int 2014;93:371-72.
30. Patenaude B, Robert Z 3rd, Scot DH. Blood—but not bleeding—at a tracheotomy site: a case of Munchausen's syndrome. ENT-Ear, Nose & Throat Journal. 2006;85:677-9.
31. Hafiz AM, Mahboob H, Jan F. Factitious gastrointestinal bleeding: a case of Munchhausen syndrome. Internet Journal of Internal Medicine. 2010:8(1).
32. Adhikary TR, Dorji T. Recurrent ear bleed with profound bilateral sensorineural hearing loss : a case of Munchausen syndrome. SAGE Open Medical Case Reports. 2021;9:1-4.
33. Yavuz ST, Cinel G, Ozcelik U, Yalcin E, Dogru D, Kiper N. Munchausen syndrome by proxy presenting as unexplained hemoptysis: a report of two cases. Europeon Respiratory Journal. 2012:40;3500.
34. Demircioglu F, Bekdas M, Goksugur SB, Gunes C, Yildirim O. A rare reason in an infant who presented with recurrent bleeding: Munchausen by proxy syndrome. Journal of Medical Cases. 2014;5:385-7.
35. Kumar R, Cherian A. Munchausen's syndrome by proxy: a case report. Indian J.Psychiat. 1994;36:195-6.
36. Gehlawat P, Gehlawat VK, Singh P, Gupta R. Munchausen syndrome by proxy: an alarming face of child abuse. Indian J Psychol Med. 2015;37:90-2.
37. Bakhurji S, Yassin SA, Abdulhameed RM. Healthy infant with bloody tears : case report and mini review of the literature. Saudi Journal of Ophthalmol. 2018;32:246-9.
38. Varalakshmi V, Doshi V, Sivalingam D, Nambi S. The story of a girl with weeping blood: Childhood depression with a rare presentation. Indian J of Psy. 2015;57:88-90.
39. Das D, Chirantan M, Meel R, Neupane, S. Crying out blood: haemolacria in a young girl, BMJ Case Rep. 2020;13:e236579.
40. Ahluwalia BK, Kurana AK, Sood S. Bloody tears. Indian Journal of Ophthalmol. 1987;35:41-3.
41. Beyazidiz E, Ozdamar Y, Beyazidiz O, Yerli H. Idiopathic bilateral bloody tearing. Case Reports in Ophthalmol Medicine. 2015;2015:692382.
42. James R, Bharadhi M, James J. Haemolacria in a 22-year-old boy. BMJ. 2018,11:e225151.
43. Das D, Kumari P, Poddar A, Laha T. Bleeding to life: a case series of hematohidrosis and hemolacria. Indian Journal of Paediatrics. 2020;87:4.
44. Agrawal S, Monishka T, Das D, Bajaj MS, Modaboyina S, , Modaboyina V. Tears of blood-a female adolescent with essential idiopathic bilateral haemolcria: case report and brief review. Tropical Doctor. 2021;50:237-40.
45. Torres JA, Otero AC, Keeling CR, Osvaldo RG. A pediatric case of haemolacria. Revista Cubana de Pediatria. 2018;90:132-40.
46. Acherjya G. Idiopathic haemalcria: a rare case report. JOM. 2019;20:106-8.
47. Rahman MS, Karim MR, Islam MM, Karim MDR. Dissociative disorders of haemolacria: series of case reports. Journal of Bangladesh College of Physicians and Surgeons. 2017;35:36.
48. Shahgoli E. A case series of hematohydrosis:a puzzling medical phenomenon. The Turkish Journal of Paediatrics. 2018;60:757-61.
49. Alsermani M, Alzahrani H, Fakih RE. Hematidrosis: a fascinating phenomenon-case study and overview of the literature. Semin Throm Hemost. 2018;44:293-95.
50. Jafar A and Ahmed A. Child who presented with facial hematohidrosis compared with published cases. Case Reports in Dermatological Medicine 2016:1-4
51. Mora E, Lucas J. Hematidrosis: blood sweat. Blood. 2013;121:1493.
52. Maglie R, Caproni M. A case of blood sweating: hematohidrosis syndrome. CMAJ. 2017;189:E1314.
53. Ucakturk A, Guindi F and Aydin M. Apparent cyclic vaginal bleeding in a child: factitious disorder. Clin Pediatr Endocrionol. 2017;26:189-92.
54. Deshpande M, India V, Kumar V, Reddy IR. Child who presented with hematohidrosis (sweating blood) with oppositional defiant disorder. Indian J of Psy 2014; 56:289-91.
55. Chatterjee SS, Mitra S. Dermatitis artefacta mimicking borderline personality disorder: sometimes, skin could be misleading. Clinical Psychopharmacology and Neuroscience. 2016;14:311-13.
56. Jefferany M, Bhattacharya G. Psychogenic purpura (Gardner-Diamond Syndrome). Prim Care Companion CNS Disord. 2015;17.
57. Palanisamy A, Subramani N, Rej S, Kothandapany S. Recurrent bleeding lip in an adolescent female - chasing the cause. Indian Journal of Dermatology; 2016;61:123-25.
58. Das S, Mohammed S, Doval N, Kartha A. Factitious disorder–a rare cause for unexplained epistaxis. Sanghai Archieves of Psychiatry. 2017;29:120-23.
59. Bakkers JTN, Crobach FSJ, Pauwels. Factitious gastrointestinal bleeding. The Journal of Nucl Med. 1985;26:666-67.

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