

Bleeding manifestations apparently unrelated to coagulation or other organic disorders: A tentative classification and diagnostic clues

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Objective: To study the features of bleeding conditions apparently not associated with vascular, platelet, or clotting dysfunctions.

Method: Conditions that may meet these criteria are: Münchhausen syndrome factitious or fictitious, suicidal or homicidal bleeding, bleeding due to self-punishment, stigmatization, the battered child syndrome, and psychogenic bleeding.

Results: The importance of these variegated conditions is not trivial in clinical practice. Differential diagnosis may be difficult and involve other specialists besides hematologists. Occasionally, invasive procedures are involved.

Discussion: The occurrence of bleeding in patients, without a clotting defect or a systemic disorder and a negative family history for bleeding represents a diagnostic challenge. A careful examination of the physical and psychological status of the patient and an appropriate evaluation of the environment in which bleeding occurs, is always needed.

Keywords: Bleeding, Psychogenic, Factitious, Suicidal, Ritual

The occurrence of bleeding manifestations is usually the result of a clotting defect, traumas, surgery, delivery, menarche, tooth extraction, etc. It may be superficial, involving skin and mucosae or deep seated, involving muscle, joints, parenchymal tissues, mesothelial cavities, gastrointestinal or urinary tracts, and the nervous system. Since bleeding affects all of the branches of medicine and surgery and may sometimes be severe or even life-threatening, it represents one of the most important signs and symptoms in clinical practice.¹⁻³

In some instances, bleeding may be factitious or fictitious, namely self-induced. The distinction between factitious or fictitious is subtle and the two forms are usually used interchangeably. Actually, fictitious means false, feigned whereas factitious means artificial as opposed to genuine or normal. In other forms, it may be suicidal, homicidal, or the expression of genitorial aggression as in the case of the battered child syndrome.

Finally, other types of non-hematological bleeding may have an atonement or self-punishing feature and a religious or mystical pathogenesis such as bleeding

from autoflagellation or stigmata. Altogether, these forms of bleeding may be grouped in terms of 'non-organic disease-related bleeding'. Needless to say that, in some instances, a hidden organic disease or still unknown coagulation defects may also be present. The purpose of the present study is to evaluate the diagnostic problems encountered by caring physicians in the diagnosis and management of these special hemorrhagic manifestations, occurring, sporadically, in patients with no apparent clotting defect and negative family for a bleeding diathesis.

Patients and classification

Personal files dealing with bleeding manifestations encountered in patients without an apparent coagulation defect, without systemic diseases, and with a negative family history for a bleeding tendency were re-evaluated. Case reports or reviews dealing with the same type of bleeding were collected from a specific time-unlimited search from PubMed using appropriate keywords.

Patient with mucocutaneous hemorrhagic manifestation due to systemic diseases or conditions (vasculitis, cryoglobulinemia, cortisone related, purpura, etc.) were excluded.

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Original papers were obtained with the help of the Pinali library of our Medical School and through the courtesy of Med Data, Sorengo, Switzerland. Cross-checking of the references listed at the end of the single papers was conducted in order to avoid omissions. On the basis of personal experience and as a result of the extensive bibliographic search, these special forms of bleeding could be classified along the following lines:

- (1) Factitious, fictitious, and Münchausen syndrome bleeding
- (2) Suicidal bleeding
- (3) Homicidal or aggression motivated bleeding
- (4) Bleeding as expression of atonement or self-punishment
- (5) Bleeding due to medications or remedies unrelated to blood coagulation
- (6) Mystic or religious bleeding manifestations
- (7) Bleeding due to ludic or circus-related performances.
- (8) Psychogenic bleeding
- (9) Bleeding due to blood bondage.

Factitious, fictitious, and Münchausen syndrome bleeding

Fictitious and factitious bleeding are commonly used interchangeably in clinical practice. As stated above, this is not fully correct since fictitious means false, feigned whereas factitious means artificial as opposed to genuine or normal. The Münchausen syndrome is a chronic and variably severe subgroup of factitious disorders characterized mainly by physical signs and symptoms.^{4,5}

Münchausen patients utilize several means to attract the physician's attention. A fictitious bleeding

manifestation is probably more frequent than originally thought. The most frequent bleeding manifestations are: fake vaginal bleeding, hemoptysis, skin hemorrhages, hematuria, and gastrointestinal bleeding.⁶⁻¹⁵ The mechanisms underlining these bleeding manifestations are often difficult to ascertain because these patients are astute, often have some medical knowledge, and tend to change doctor or hospital from time to time. Sometimes, they fake medical records. To supply a balanced idea of the complexity of bleeding in Münchausen syndrome a few reported cases are gathered in Table 1. In most of these cases, the correct diagnosis was reached after an extensive evaluation of the patients, evaluation which included hospital admissions, a variety of diagnostic tests, including invasive procedures like biopsies.⁵

Hemoptysis is one of the most frequent bleeding manifestations in Münchausen patients.¹¹ These patients usually undergo several procedures (chest X-rays, CAT, bronchoscopy etc.) at a tremendous cost for the Health System, before the correct diagnosis is reached. In the case of hemoptysis occurring in concomitance with the menstrual cycle, catamenial bleeding has to be considered.

Bleeding has been involved also in the Münchausen syndrome by proxy or in Polle's syndrome. In this case, the fake or feigned bleeding involves a third party, usually a son or a daughter.¹² The mother intends to draw attention by utilizing the factitious manifestation induced by her on someone else.¹⁶

Suicidal bleeding

Suicide due to cutting of the wrist veins is a well-known event. It was more frequent in the past. In

Table 1 Bleeding manifestations seen in patients with Münchausen's syndrome

Author/year	Gender/age	Site and type of bleeding manifestation	Postulated mechanism for bleeding and comments	
Bakkers <i>et al.</i> ⁶	27, F	Gastrointestinal	Auto-induced enemas of blood samples deceitfully obtained	
Michalowski ⁷	24, F	Cheilorrhagia	Local scratches and piercings	
Karnik <i>et al.</i> ⁸	18, F	Superficial multiple sites	Patient used to inject air in subcutaneous tissues	
Lazarus and Kozinn ⁹	24, M	Hematuria	Ingestion of rat poison (superwarfarin)	
Bakri and Jabbar ¹⁰	20, F	Menorrhagia	Fictitious ingestion of oral anticoagulants	
Baktari <i>et al.</i> ¹¹	36, M	Hemoptysis	Mechanism of bleeding undetected (paper contains a list of previously reported cases)	
Bennett <i>et al.</i> ¹²	29, F	Otorrhagia in son	The mother spat blood in the child's ears. Example of Münchausen syndrome by proxy	
Petersen and Barthels ¹³	Variable 9, F 5, M	Several manifestations mainly mucocutaneous	Factitious intake of Phenprocoumon (the paper reports on 15 patients)	
Girolami <i>et al.</i> ¹⁴	Case 1	19, F	Skin and subcutaneous hemorrhagic nodules	Local injection of perfumes
Case 2	20, F	Nipples and areolar areas	Smearing and/or injection of menstrual blood in nipples and areolas (patient underwent bilateral mastectomy on suspicion of cancer)	
Saed <i>et al.</i> ¹⁵	22, F	Hemoptysis	Mechanism undetected. Possibly catamenial in origin	

The list is not complete but only indicative of the type of faked bleeding and of its underlying mechanism manifested or adopted by these patients.

the past decade it has been substituted as a frequent case of suicidal attempt by the ingestion of hypnotic or similar medications. There are also rare cases of suicidal attempt by the use of excessive coumarin medications. These include not only the compounds used for normal anticoagulant but also the more toxic compounds used as rodenticides or superwarfarin.^{17–19} In the latter case, it is important to know that no Coumadin or warfarin is found in the blood of these patients, but only the Coumadin-like substance or superwarfarins responsible for the bleeding, for example, ‘Brodifacoum’. Prothrombin time in these patients is prolonged but plasma warfarin levels are normal. Specific assays are needed to detect the culprit.¹⁸

It is worth noting that patients who have taken these superwarfarins may present massive bleeding and venous thrombosis at the same time due to a sharp decrease in Protein C and Protein S levels besides the fall of factors II, VII, IX, and X.²⁰

Homicidal or aggression motivated bleeding

Attempted or successful homicides due to administration of high doses of anticoagulants, usually superwarfarins, have been described.²¹

Other aspects of aggression-related bleeding are represented by the ‘battered child syndrome’ and the ‘shaken baby syndrome’.^{22–24} The main clinical features of the ‘battered child syndrome’ are multiple bruises and hematomas associated often with bone fractures. The clinical picture, especially if no fractures are present, resembles a baby with a congenital or acquired coagulation disorder.^{1,25} This is complicated by the fact that the parent or parents accompanying the child tend to deny any responsibility for the event and claim that the child has fallen accidentally. It is important to find out the plausibility between the type and site of the hemorrhagic lesions and the reported trauma.²⁴ This seldom occurs. The ‘shaken baby syndrome’ is characterized by conjunctival and/or retinal hemorrhages with occasional swelling of face tissues. Neurological signs compatible with subdural hematoma and fracture of the long bones may also be present. The lesions are due to violent shaking of the baby, which causes a violent ‘acceleration–deceleration’ mechanism. The infant, due to the disproportion between the size and the weight of the head and that of the body and with still weak neck muscles, is particularly prone to have the skull structures severely damaged.²³

Under this heading, vampirism could be also included. Vampirism is a rare condition characterized by an urge and impulse to suck blood usually obtained by biting another person.²⁶

The condition had, and still has even today, literary, anthropological, medical, and psychiatric significances.

It was maintained to be frequent in the Balkan states and this was probably a reflection of the literary work of Stoker who wrote *Dracula*,²⁷ first published in London in 1897.

Several medical tentative explanations have been proposed as the underlying cause for vampirism. Rabies, Pellagra, and Porphyria erythropoietica were the most widely used together with several psychiatric disorders.^{28–31} Today, it is considered a psychiatric disorder not without potential organic implications. Auto-vampirism has been reported, for example, as a cause of anemia.³² Vampirism is one of the most popular myths among several civilizations.³⁰ It could be envisaged as the archaic struggle between life and death, since blood is usually identified with ‘life’ and the lack of blood with ‘death’.^{26–30}

Bleeding as an expression of atonement or punishment

The typical example of this type of bleeding is autoflagellation. The procedure is still widely used during procession or other religious gatherings in several Mediterranean countries. The typical hemorrhagic lesions are represented by ecchymotic streaks involving mainly the shoulders and back. Deep hematomas are rare but secondary infections of skin lesions are common.

Strictly associated with this form of bleeding is the *Lasthénie de Ferjol syndrome*.³³ The heroine of the book by Jules Amédée Barbey d’Aureville, ‘*Une histoire sans nom*’, was desperate and shameful for an unwanted pregnancy due to a sexual violence and for the continuous reproaches she received from her mother. As a reaction to this predicament, she wanted to punish herself by sticking needles in her precordial area. These needles caused repeated staining of her blouses and, with time, severe anemia. ‘*Lasthénie killed herself slowly, killed herself, every day a little more with needles*’ wrote Barbey d’Aureville. The condition once it reaches this extreme result could be also considered as part of suicidal bleeding. Fortunately, usually this is not the case and therefore the syndrome is also known as ‘*Anemia by self-injury*’.³³

Bleeding due to ingestion of medications or remedies apparently unrelated to blood coagulation

Several antiplastic agents are associated with bleeding due to platelet decrease. Acute severe intoxication with LSD has been reported to be associated with diffuse bleeding of unknown cause.³⁴ Herbal supplement use may be associated with bleeding, particularly frequent with the ingestion of preparations containing *Ginkgo biloba*.^{35,36} This is a widely used herbal remedy not only in the treatment of memory loss, vertigo, tinnitus,

cerebrovascular insufficiency but also in asthma and allergies.

The mechanism of action is unknown but an effect on platelet-vascular function has been suspected since patients often show a prolonged bleeding time. Since the majority of patients who show bleeding are on anticoagulant or antiplatelet medications, it is likely that Ginkgo biloba causes a potentiating effect of these compounds.³⁷

Bleeding due to religious or mystic significance
Stigmata are the most frequent manifestations.³⁸ The scientific approach to this condition is limited to the exclusion of the existence of a hemorrhagic diathesis in the person who shows stigmata or other peculiar bleeding manifestations. Some of these bleeding manifestations may be self-induced and be part of the Münchausen syndrome.³⁹ Reported bleeding from religious images or statues may also be grouped in this category.

The ritual and periodic transformation of 'blood' contained in ampoules and vials from the gel to the fluid state can be also included in this chapter. Scientific proof is difficult to obtain since the possibility of control is denied or limited. The event has to be accepted only as an act of faith, even though several theoretical explanations for 'blood' liquefaction have been advanced. Thixotropy, namely the capability of certain gels to liquefy after mechanical stress, has been proposed as an explanation.^{40,41} Another explanation is based on the possibility that reliquaries contain spermaceti, ether, and red dye from the roots of the alkanet plant. This mixture is solid at 10°C but melts when held in the hand due to body heat.⁴² The most widely known example of this blood liquefaction is that of St Januarius in Naples. St Januarius was a Bishop and a martyr, beheaded by Emperor Diocletian. His blood is contained in a sealed glass vial in Naples's cathedral. The 'blood' has the ability to liquefy when the vial is turned up and down for a while.⁴⁰ There are other similar examples of 'blood' liquefaction. For example, in Catholic religion, a blood relic of St Pantaleon of Nicomedia, a physician, is reported to undergo periodic liquefaction in Madrid. The same is true for St Laurence's 'blood' in Amaseno (Italy). These Saints were all martyrs brought to death for their faith during the persecutions carried out by Roman Emperors during the third century after Christ.

Bleeding due to ludic or circus-related performances

The typical expression of this condition is represented by the 'sword swallows'. Massive gastrointestinal bleeding is, together with perforation,⁴³ the most important complication of this procedure.

Psychogenic bleeding

The most important entity on this front is represented by Psychogenic purpura also known as Gardner–Diamond syndrome or autoerythrocyte sensitization syndrome.^{44–46} This is a condition that affects mainly women in fertile age. It is characterized by purpuric lesions that involve mainly the extremities. The manifestation is sporadic but often recurrent in concomitance with stressful conditions, surgery, and traumas. Sometimes, the purpuric lesion may be accompanied by local pain and by nervousness, malaise, headache, dysphagia, and paresthesias. These patients often present depression, feelings of hostility, obsessive-compulsive disorders, and trait of hysterical or conversion disorders. The lesions can be triggered in some patients but not in all, by the intracutaneous injection of the patient's own erythrocytes. No sure alteration of blood coagulation has been demonstrated. Ratnoff in an excellent review in 1989⁴⁴ concluded that the condition is an unsolved dilemma. Today, the condition is still unexplained even though, hormonal imbalances and increased cutaneous fibrinolytic activities have been proposed as possible causative factors.

In this chapter, we could also include the cases of bloody tears or hemolacria and bloody sweating or hematidrosis of non-organic nature.^{46,47,51} In agreement with this interpretation, Ho *et al.*⁴⁸ in their extensive study on the subject, consider this form of bloody tears to be caused by hysteria stigmatization. Hematidrosis or bloody sweating is a rare condition which may have a psychogenic cause once other causes such as systemic disease, vicarious menstruations, and excessive exertion have been ruled out.^{47,51} The differential diagnosis of these psychogenic forms may be difficult. The latter conditions usually affect females with personality pattern or conversion disorders.

Bleeding due to blood bondage

Blood bondage also called blood covenant is a ritual of friendly and mutual commitment between two persons. Apparently, it was common in the Medieval times and among pre-Colombian American populations.⁵² It involves usually two men, who by mixing their blood became blood brothers. The bleeding is usually provoked by a cut with a knife or stilet in the arm. The cut is simultaneously produced in the two persons who, immediately after, mix their blood by pressing together the two parts of the body with the bleeding wounds. Occasionally, blood bondage may have the significance of adhesion to a sect or secret organization.

Discussion and conclusion

No information is available in the literature about the prevalence of this type of bleeding in the general

population of bleeders. Nonetheless the problem exists and, judging from the ever increasing number of bleeding as part of the Münchhausen syndrome, may not be trivial^{13,14} and has to be faced.^{1,2}

These peculiar bleeding manifestations, unless clearly justified by the environmental or personal setting, are usually first confronted by the general practitioner or the internist. Only subsequently are they brought to the attention of the hematologist and finally to the coagulation expert.^{14,25} Discrepancies arise when the laboratory results fail to show any abnormality and the presence of systemic diseases such as vasculitis, crioglobulinemia, etc. have been excluded. At this point, two routes are open for the caring physician, namely request more complicated or unusual clotting tests (antiplasmin deficiency, local fibrinolysis studies, platelet function studies, FXIII deficiency etc.) or, two, investigate carefully the setting in which bleeding occurs. Needless to say the two approaches may be concomitant.

This is particularly true for the Munchausen syndrome and stigmata. The latter may have a religious meaning for the believer but have limited significance for the clinician. On the contrary, the bleeding manifestations occurring in Munchausen syndrome or in the battered child syndrome represent difficult diagnostic problems and are frequently potential causes of medico-legal litigation. In the evaluation of battered child syndrome, it is critical to exclude an underlying coagulation defect and to gather an accurate psychological history of all family members.

Several hemorrhagic manifestations have been described in the Munchausen syndrome or in other cases of factitious bleeding. The selected cases reported in Table 1 clearly indicate the variety of the faked bleeding manifestations. The sites of this factitious bleeding may trigger an extensive and expensive investigation. Sometimes, it may require hospital admission. The type and site of bleeding manifestations have to be carefully evaluated. The family setting and the psychological profile of the patient have to be thoroughly investigated.

In the case of cutaneous manifestation it is important to notice the areas of the body involved. Usually these areas are within the reach of the arms of the patients (legs, arms themselves, anterior part of the body, and face). Furthermore, the occurrence of bleeding is never witnessed by a family member. It always occurs when the patient is alone. Family members should cooperate in controlling the patient for the possession of needles, syringes, and other tools. The diagnosis of bleeding due to autoflagellation is easier, but when the wounds are complicated by infection and the self-inflicted lesion denied, diagnosis may be difficult. Usually, the approach to a bleeding patient is based on balanced synthesis of

clinical and laboratory findings.^{1-3,49} This is not the case in this type of bleeding. The approach, once clotting defects and systemic diseases have been excluded, must be based only on clinical grounds. The physician's experience and alertness play a fundamental role, but this clinical approach has to include a careful evaluation of the psychology of the patient, of her or his parents and of the environment or setting in which the bleeding manifests itself. When dealing with bleeding manifestations *sine natura*, namely without an underlying coagulation defect, the possibility of cheating or faking on the part of the patient or of his on her parents is always to be kept in mind. The physician is justified to rule out this suspect only after a careful evaluation of the patient and of his on her family setting and environment. For this purpose, a strict cooperation with psychologists and psychiatrists is often necessary. Finally, one has to keep always in mind the potential limitations of laboratory tests now in use and the possibility that still unknown coagulation or platelet defects might contribute to these fictitious bleeding manifestations.^{49,50}

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