RECURRENT EXTRAGENITAL HAEMORRHAGES

BY

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To the occurrence at regular intervals of extragenital haemorrhages from one or other part of the body at or near the time of the menstrual period the name "vicarious menstruation" has been given. In the majority of the cases described in the literature such a designation is not strictly accurate as there is a lack of convincing evidence to show that the abnormal bleeding is, in reality, in the nature of a menstrual discharge. With a few exceptions they can merely be classified as cases showing recurrent extragenital haemorrhage with an indefinite relationship to the menstrual cycle. But, although the occurrence of true vicarious menstruation is so rare, the old name seems to have come to stay and is still handed down from one generation of students to another.

Two varieties of vicarious menstruation are described. In some instances the haemorrhage appears to take the place of the normal uterine menstrual loss, which is altogether absent. In other cases menstruation occurs, with in addition some form of extragenital bleeding. The former variety is known as substitutional while the latter is called supplementary menstrual haemorrhage.

Review of the Literature

The condition is a relatively rare one. "This extremely rare abnormality," says Bethel Solomons (1934), and Novak (1931) credits Lawson Tait with the remark, "I do not absolutely deny that there is any such thing as vicarious menstruation, but I do deny the propriety of examiners at the University of London, or anywhere else, asking the commonest cause of epistaxis and receiving the answer of vicarious menstruation with approval!" Novak (1931) quotes an extensive study of the literature made by Roth, who traced 225 cases since 1870. In his series the sources of the haemorrhages were: nose, 30 per cent.; nipples, stomach, mouth, and throat, 4 to 5 per cent.; skin and fistulae, 10 per cent.; lungs, 8 per cent.; bladder, eye, ear, intestines, and vagina, $3\frac{1}{2}$ to 4 $\frac{1}{2}$ per cent. Of his cases in 25 per cent. the bleeding was purely vicarious—that is, substitutional; in 75 per cent. supplementary to the normal uterine loss.

Other authorities disagree with these findings. Bethel Solomons (1934) regards haemoptysis, haematemeses, melaena, epistaxis, and purpura as being the most common manifestations, but the general consensus of opinion seems to place the sources of the haemorrhage in order of frequency as follows:

1. *Nasal Mucous Membrane*.—The haemorrhage may be quite severe or only very slight; but in either event it may persist throughout the normal period. Apparently no one has yet explained why the nasal mucous membrane should so often be the source of these bleedings, but Novak (1931) suggests that the cause may be closely related to the occurrence of the so-called nasal dysmenorrhoea.

2. *Stomach*.—Usually only slight bleeding.

3. *Intestine*.—Generally slight and appearing as blood in the stools during the menses.

4. *Lungs*.—Haemorrhages may have fatal results.

5. *Breasts*.—As a rule merely a sero-sanguineous discharge at the menses, but there may be definite blood.

6. *Skin*.—Haemorrhage from the skin occurs as "bloody sweat." Erythema of local patches of skin develops and is followed by oozing of pure blood, or there may be a purpuric rash only.

7. *Lips*.

8. *Eyes and Eyelids*.—Haemorrhage into the retina with each period has been recorded by Huizinge, Powell, and others. Claiborne (Novak, 1931) has reported one case of vicarious haemorrhage from the lower lids.

9. *Naevi*.

10. *Kidneys*.

11. *Old cicatrices*.

12. *Abdominal fistulae*.

13. *Umbilicus*.—A case is reported by Gardner (1898). For three months in succession his patient bled from the umbilicus at the menses for twenty-four to forty-eight hours.

Other rarer sites are teeth, gums, varicose ulcers, amputation stumps, external auditory meatus, nails, and haemorrhoids.

Problem of Causation

There is one common factor in most of the reported cases—namely, that uterine menstruation has been suppressed or less than normal. In the majority of cases irregular amenorrhoea has been the rule. In spite of much speculation no satisfactory explanation has been put forward. Most of the theories are based on the assumption that menstruation is not a local but a systemic process. Barnes (Novak, 1931), in a review of the explanations adduced, submits the theories:

1. That it is caused by plethora.

2. That it is the result of a weakened condition of the tissues, perhaps hereditary.

3. That it is due to an abnormality in the structure of the blood vessels.

More recently W. H. Condit (1916), in the *American Journal of Obstetrics*, laid emphasis upon changes in the blood pressure at the time of menstruation. These changes may be summarized briefly as being a rise in systolic pressure prior to and during the early part of menstruation and a diastolic rise during the period. The pulse pressure is above average during the flow. His evidence is unconvincing, however. A more reasonable explanation than any of the foregoing is given by Novak (1931), who suggests that in certain cases there is present in the various tissues an abnormal sensitivity to the action of the blood-borne ovarian hormone, with a result analogous to its action on the uterine endometrium. That, at least, sounds logical, more so than the idea that while the tissues themselves are normal an excess of ovarian hormone derived from supernumerary ovaries or accessory ovarian tissue, such as is found in the broad and ovarian ligaments, may precipitate the vicarious haemorrhages.

Menstruation and the Nervous System

In a proportion of cases there is a very definite relation between epilepsy and menstruation. In so-called hystero-epilepsy the epileptic seizures come on a day or two before or at the onset of the period in the great majority of cases. In the intermenstrual interval these hystero-epileptics have no seizures. On the other hand, menstruation often brings on hysterical attacks in girls and women already predisposed. In many such cases there is dysmenorrhoea of the spasmodic variety. Dipgen and Schröder (1906), in a series of seventy-five hysterical women, noticed that menstruation began at an unusually advanced age and that it was abnormally scanty. This they suggest as indicating defective development of the reproductive organs.

D. M. Greig (1932) undertook an extensive review of the subject of recurrent bleeding from the eyes. He quotes many recorded cases which serve to show that the hysterical state is more often the exciting cause than are puberal readjustments and menstrual irregularities.

Although hysterical fits may be more apt to occur at menstruation it is the instability of the nervous system and the hysterical state that are more to blame than the local utero-ovarian upset. He emphasizes the significance of the fact that so many of the women who exhibited the phenomenon of recurrent ocular bleeding were potential epileptics. He concludes that these ocular haemorrhages, though a few were connected with the menses and a few brought on by postural effects, were for the most part not connected in any way with menstruation, the cause lying in a central psychic excitation and not in a local mechanical one.

Case Report

The case which is the basis for these general remarks was that of a young girl aged 18, a hotel waitress, to whom I was first called on April 17th, 1936, as she was said to be having "fits" and "bleeding from the eyes." I arrived just after she had recovered from an attack, and was told that since August, 1935, she had suffered from recurrent attacks of bleeding from the nose, mouth, eyes, and ears accompanied by "fits."

HISTORY

She first menstruated at the age of 16. Previously she had had "catarrh of the womb" (? discharge), which was untreated. At first her periods were irregular at six-week to eight-week intervals, with loss for three to four days. Later they became stabilized, lasting for four to six days every twenty-eight days, with moderate loss and spasmodic dysmenorrhoea. In August, 1935, she had an attack of "? sunstroke," when her face was very red and she had marked infra-orbital oedema. She was ill for two months, and had amenorrhoea. On her recovery the periods altered, to last for three days at twenty-eight to fifty-six-day intervals, with a very slight loss. It was at that time (October, 1935) that the extragenital haemorrhages from the nose, mouth, ears, and eyes—chiefly the latter—began. The bleeding had been seen by many people, who said that from the eyes "drops of blood ooze out." Up to April, 1936, these haemorrhages coincided with the times for her periods, and she had the normal loss at the same time on two occasions.

In November, 1935, she was operated on at Ulverston and had the "womb stretched"—presumably a dilatation and curettage—and thereafter had only menstruated *per vias naturales* twice, in February and in April, 1936. The other haemorrhages continued at the expected time of menstruation. A point of interest is that while she was in hospital awaiting operation she saw a girl in the ward having a hysterio-epileptic attack, which frightened her and made her hope she would not have similar seizures. In February, 1936, there was normal uterine loss, with no extragenital haemorrhages, and two "fits." In March, 1936, there was amenorrhoea with bleeding from the eyes and a slight haematemesis. In April she was seen on the third day of her period. There had been numerous "fits" and frequent bleeding from the eyes, nose, and ears before the onset of her period on April 14th. She said she could tell when the eyes were going to bleed as the haemorrhage was preceded by an intense bilateral headache.

She had had chorea in 1930 and rheumatic fever with a "diseased heart" (uncorroborated on examination). There was no history of epilepsy. The patient came from a mining village, and had had a very unpleasant home life. Her mother was dead, and her father and one other sister were at home. The father was more or less a sexual pervert, and had often attempted intercourse with his daughters.

EXAMINATION

She was a well-built, well-nourished girl, but rather anaemic. Quite alert mentally, but during questioning she had another attack while sitting in her chair. She went flaccid, her head drooped as if she were asleep, and ordinary tears poured down her cheeks from both eyes. Pulse and respiration were unaltered, and the corneal reflex was present. The attack lasted about forty seconds, and thereafter she

became quite normal again. Detailed examination revealed no abnormality. Heart, lungs, abdomen, mouth, throat, and eyes were all apparently normal. The thyroid was not enlarged. Blood pressure was 126/82 mm. Hg; no abnormal constituents were discovered in the urine.

Discussion

The problem presented by this case seemed extremely complex if the story were really true. First, the fits might be either functional—that is, hysterical—or a true petit mal. Both of these conditions, as we have seen, are quite common in relation to menstruation in women of the nervous or epileptic type. Secondly, the bleeding from the mucous membranes might have two explanations: (a) there might be some very minute self-inflicted injury, or (b) it might be a true example of vicarious menstruation. So that the problem presented was to determine, if possible, what part was played by any functional lesion of the nervous system, and secondly, whether local treatment or gynaecological therapy, by hormones, could offer any prospect of cure to the patient.

At first I regarded the "fits" as being in the nature of true minor epilepsy, but subsequently revised my opinion in favour of a hysterical basis. Operation seemed to be contraindicated—dilatation and curettage having been done without success—as it was likely that any further operative trauma might increase the nervous reaction of the patient.

Treatment and Progress

The treatment given consisted of calcium and sodium lactate 10 grains three times a day, with phenobarbitone tablets 1/2 grain morning and night, and injections of "plurigland" (Oxo) once a week, subsequently altered to antuitrin S daily on the advice of Professor Leyland Robinson, for three days before the period and daily throughout.

April 26th, 1936.—Period due in fourteen days. To-day bled from both eyes. Seen by me in surgery with dried blood on lids and cheeks. No sign of injury.

April 30th.—Recurrence of haemorrhage from both eyes. Conjunctivae flushed; cornea dull; severe photophobia.

May 7th.—Period due on the 9th; injection of 1 c.cm. antuitrin S given.

May 8th.—Injection of antuitrin S.

May 9th.—Injection of antuitrin S. Patient dreads injections, and had a "fit" in the surgery while the syringe was being filled. Not a true petit mal—definitely hysterical.

May 10th.—Period began previous night; haematemesis.

May 11th.—Profuse haematemesis and frequent haemorrhages from both eyes and the right ear, definitely confirmed by three witnesses.

May 12th.—Considerable haemorrhage from eyes and from the left ear; severe haematemesis; period ceasing. Admitted to Westmorland County Hospital.

May 13th.—During the night following admission she had slight haemorrhage from both eyes and mouth; blood seen coming from the eyes during sleep; slight recurrence from eyes at 3 p.m.; blood pressure 120/80 mm. Hg; red blood cells, 5,260,000; white blood cells, 6,400 per c.mm. (House-surgeon's count.)

May 14th.—Slight haemorrhage from eyes seen at 10 p.m., 10.30 p.m., and 4 a.m.

May 15th.—Bled from eyes several times and from mouth and right ear.

May 16th.—Blood from eyes and right ear.

May 17th.—No further haemorrhages.

May 19th.—Discharged at own request to her home in West Cumberland.

July 1st.—Admitted to Royal Southern Hospital, Liverpool, under Professor Leyland Robinson. On day of admission bled from inner canthus of both eyes.

July 2nd.—Epistaxis; haemorrhage from ears and eyes.

July 4th.—Bleeding from ears, eyes, nose, and umbilicus.

July 5th.—Slight haematemesis; period began.

July 12th.—Has had haemorrhages off and on from eyes, ears, nose, and umbilicus; period ended.

July 13th.—Seen by physician; no evidence of organic lesion of central nervous system; heart sounds normal; blood pressure 115/80 mm. Hg.

July 14th.—Blood examination: red blood cells 4,400,000, diameter 7.17μ , haemoglobin 74 per cent., colour index 0.84, bleeding time normal; white blood cells 7,750 per c.mm.; differential count: polymorphs 69 per cent., eosinophils 6 per cent., monocytes 1 per cent., lymphocytes 24 per cent. (large 10 per cent., small 14 per cent.).

July 16th.—Blood calcium 10.03 mg. per 100 c.cm.

July 22nd.—Seen by ear-nose-and-throat surgeon, who reported no source of bleeding manifest in the auditory apparatus.

July 24th.—Seen by ophthalmologist. Vision normal in both eyes; bluish sclerotics; fundus normal.

July 28th.—Discharged.

August 15th.—Seen at surgery. Feels and looks well. Period on August 1st as expected; no accompanying haemorrhages.

Thereafter lost sight of as she left this district.

Conclusions

Into what category can we fit this case? The salient features are a history of previous "rheumatism of the nervous system"; with a degrading home life and with a half-formed dread of developing hystero-epilepsy in a girl who, after a definite illness, exhibits hysterical attacks at the time of menstruation, accompanied at times by extragenital haemorrhages. Such a picture does not altogether correspond with one's idea of true vicarious menstruation—that is, substitutional haemorrhage. The sequence of events is not definite enough—sometimes she had "fits," sometimes amenorrhoea, sometimes normal uterine loss, and sometimes extragenital haemorrhages, and sometimes a combination of these phenomena. Nor does it fit in satisfactorily, though it is closer to it, with one's ideas of the supplemental variety of vicarious menstruation, for the haemorrhages did not always correspond with the menses.

A study of the facts of this case offers no explanation as to the exact mechanism of the haemorrhages, except of a rather negative kind. In this case they did not seem to result from plethora nor changes in the blood pressure. Nor does her past menstrual history suggest that defective development of the pelvic organs was a factor in their causation. Apart from a tentative and non-committal "vasomotor instability" there seems no obvious explanation. Certainly self-inflicted trauma can be altogether ruled out—the bleeding was seen by many witnesses, including sceptical sisters, nurses, and house-surgeons.

There is, however, no doubt in my mind but that the "fits" were purely hysterical in character. An old choreic is a notoriously emotional individual at best, and this girl's attention would seem, by a chain of circumstances, to have been focused on her menstrual habits and irregularities, with a resultant mental disturbance.

My thanks are due to Professor Leyland Robinson for his interest in and help with this case.

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THE FRIEDMAN TEST IN HYDATIDIFORM MOLE AND CHORION EPITHELIOMA

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The following four cases in which the forty-eight-hour Friedman test was used show the value of the test so clearly that they seem worth recording. The test was used (1) to establish the presence of a hydatidiform mole or a chorion epithelioma, (2) as a criterion of the complete removal of a mole or the presence of a subsequent chorion epithelioma, and (3) as evidence of the absence of secondary deposits after hysterectomy for chorion epithelioma.

Case I

A patient, aged 29 years, was admitted on January 6th, 1934, for an incomplete abortion. The menstrual periods had been regular, the last being on October 29th, 1933. She started bleeding on December 21st, and the loss had continued off and on until admission; after admission clots were passed at times. On January 16th, 1934, the os was dilated and two pieces of what resembled placental tissue were removed. On the 22nd products of conception resembling a vesicular mole were discharged. Histological examination showed a hydatidiform mole with considerable degeneration. On January 30th the Friedman test was positive. She was sent home on February 10th to attend as an out-patient for further tests. On March 6th the reaction was positive and again on March 26th, when she was readmitted for vaginal haemorrhage. Panhysterectomy was performed two days later on the strength of the Friedman test having remained positive for over two months after the passing of the mole. There was a small tumour in the uterine wall at the fundus, which proved histologically to be a chorion epithelioma. On April 15th the test was positive, and on April 23rd weakly positive. The patient was discharged on May 17th, and further tests as an out-patient on July 3rd, August 1st, and August 27th were all negative.

Case II

A patient, aged 27 years, was admitted on January 11th, 1934, for an incomplete abortion. The periods had been regular but profuse, the last being on September 12th, 1933. In October, 1933, a brownish discharge began and persisted. Two weeks before admission she started to lose blood, at times profusely. On January 27th, 1934, the cervix was dilated and the products of conception removed. These resembled a hydatidiform mole, and histologically very degenerated and oedematous chorionic villi were seen. On January 31st a vesicular mole was passed, and on February 16th the patient was discharged. On February 26th, March 26th, and May 24th the Friedman test was negative. On September 2nd the patient reappeared complaining of a brownish discharge; the test was negative. On December 27th the test was positive, with a history of nine weeks' amenorrhoea. The patient has since been delivered of a full-term healthy child.

Case III

A patient, aged 25 years, was admitted on April 12th, 1934, for flooding after a two-months abortion six weeks previously. On April 15th the uterus was explored and a friable mass removed; histologically this was a chorion epithelioma. On the 16th the Friedman test was positive. Panhysterectomy was performed on the 23rd. On May 1st the test was weakly positive, and it was completely negative on May 22nd, June 25th, July 3rd, and November 12th, 1934.