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Mental Symptoms and Personality Structure in Autoerythrocyte Sensitization Syndrome

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Introduction

The syndrome of autoerythrocyte sensitization is characterized by painful ecchymotic lesions of the skin that develop in a very stereotypic manner. Onset occurs in a palm-sized skin region, usually on the limbs, with a stinging and burning sensation. After varying lengths of time, in general 2–6 hours, the skin becomes red, hot, oedematous and tender. This inflammatory phase may last anything up to twenty-four hours. Over the next few days the rubescent heated infiltration turns into an ecchymosis which spreads annularly at the same time as the tenderness and swelling diminish. Originally described by Gardner and Diamond (1955), thirty-one cases of the syndrome have now been reported. Up to the present all reported cases have come from North America and Australia and been confined to women with an age at onset of between 15 and 66 years.

Gardner and Diamond first noted that intracutaneous injection of red cell stroma could produce the lesions, and in our opinion this so-called erythrocyte test should be an obligatory diagnostic criterion. Most patients have had their manifestations for years and undergone very comprehensive medical examinations which have failed to disclose any haematological abnormalities or coagulation disturbances, nor has the erythrocyte sensitivity been discovered. Apart from the typical tender skin haematomata, a high proportion of these patients exhibited mental and neurological symptoms, abdominal pains, menometrorrhagia, gastrointestinal haemorrhages, haematuria and epistaxis. In remarkably many cases the onset of the disease is preceded by minor traumata or postoperative erythrocyte extravasation into the tissues.

Review of the Literature

The occurrence of mental insufficiency symptoms in autoerythrocyte sensitization syndrome was initially pointed out by Ratnoff and Agle (1961) and Agle and Ratnoff (1962). Several of their patients exhibited so-called hysterical manifestations such as aphonia, paralysis, paraesthesia, convulsions and attacks of syncope. All suffered from functional autonomic disturbances including periodic vomiting, irritable colon syndrome and recurrent headache. Moreover it was noteworthy that their relations to other members of the family were strained and also that they were maladapted to the stresses of life. Purpuric lesions were ascribed to emotional stress. Agle and Ratnoff also maintained that so-called masochistic character traits were frequent, adducing in support of this the high incidence of sadomasochistic relationships and an addiction to surgical procedures, particularly those involving the sexual organs. It was also noted that such patients often adopt an attitude of 'grin and carry on', not only in the face of their illness but to any adversity, suggestive of actual enjoyment of the hardship.

A subsequent paper (Agle and Wasman, 1967) similarly drew attention to the striking uniformity of the psychiatric findings as the clinical picture became more variegated. The authors stressed the occurrence of strong dependency needs often manifested in a passive aggressive manner, feeling of helplessness, incompetency and need to be protected. These needs were often expressed in a selfish egocentric demanding fashion. All patients were thought to be naïve and childlike in their interpersonal relationships. Such a description of these patients obviously justifies their being classified as psychoinfantile (Lindberg, 1950, 1953).

Papers published prior to 1961 merely make passing reference to manifestations of psychiatric value. Thus, Henstell and Kligerman (1957) reported a case in which the patient had acute episodes of nausea with unproductive retching at irregular intervals and episodic pain in the abdomen which was not related to the nausea. McCay (1960) described a case with protracted hiccups, headache, malaise, generalized weakness, nausea and dizziness. This condition developed following a mild head trauma whereupon transient hemiparesis and hemihypoaesthesia also supervened. Cerebral thrombosis was presumed to be the causative factor, although neurological manifestations were absent and the spinal tap and carotid arteriography were normal. Reed and Firkin (1957) described a case in which there was severe headache, temporarily reduced visual acuity, hemiparesis and episodic vomiting. The CSF was clear and under normal tension, arteriograms and ventriculograms were normal and the condition was attributed to intracerebral haemorrhage. Two of Gardner's and Diamond's (1955) original four patients had transient hemiparesis which was presumed to be associated with cerebral haemorrhage (Case 1 and 4). In addition Case 1 on one occasion had disturbed vision in the left eye and was found to have a dilated left pupil. During several years Case 2 had unexplained attacks of syncope. Case 3 had abdominal distress and much vomiting after physical trauma, which necessitated hospitalization and parenteral fluid therapy on two occasions. Case 4 had many attacks of nausea and vomiting.

Thus functional disturbances of various types were noted in the early cases too, and hence they comply well with the observations of Agle and Ratnoff. In the light of the high incidence of hysteriform mechanisms in Agle's and Ratnoff's series it is tempting to question the proposed diagnoses of cerebral thrombosis (McCay, 1960) and cerebral haemorrhage (Reed and Firkin, 1957; Gardner and Diamond, 1955). In none of these cases is the clinical picture entirely convincing, and the possibility cannot be ruled out that it actually was a matter of hysteriform symptoms. That is not to say, however, that such manifestations cannot often have an organic cerebral background.

McDuffie and McGuire (1965) published a series of five cases in whose histories paralysis and syncope also commonly occurred. All these patients had been subjected to the MMPI questionnaire which revealed, among other things, the probable presence of the following personality traits: they frequently had mild somatic signs, especially in stressful situations; they had considerable difficulties in handling feelings of aggression and hostility; they were often extravert and sociable but at the same time difficult, irritable and demanding; they commonly displayed hypochondriacal and hysteriform attitudes. Essentially, therefore, the descriptions of McDuffie and McGuire agree well with those of Ratnoff and Agle.

A serious problem in the discussion about the pathogenesis of the disorder is the possibility of self-inflicted bruising. One case exhibiting characteristic features of autoerythrocyte sensitization syndrome has been described which on closer examination turned out to be factitious purpura (Davidson, 1964). In two other cases, the patients were actually seen striking themselves (Agle and Ratnoff, 1962; McDuffie and McGuire, 1965). However, the possibility of self-induced artifacts due to malingering or dissociative episodes has been ruled out in various ways in several investigations. Thus Gottlieb *et al.* (1957) described a patient who was placed in a cast to preclude self-trauma, and it was demonstrated that she continued to develop lesions beneath the cast.

Groch *et al.* (1966), by extensive double-blind testing of a patient and control subjects, revealed in autoerythrocyte sensitization syndrome a reactivity against erythrocytes as well as against phosphatidyl serine which is a component of erythrocyte stroma. In their studies particular effort was made to eliminate possibilities of suggestion, trauma, or bias in the development of lesions. The patients were under close observation in a research unit during the progressive development of the ecchymoses, suggesting that these lesions were not self-induced. The developing lesions were biopsied three, twelve and eighteen hours following the intradermal administration of phosphatidyl serine. At twelve hours there was a massive increase in the number of perivascular poly-

morphonuclear cells and, in addition, these cells diffusely infiltrated the upper dermis giving the appearance of a cellulitis. Endothelial cells and the surrounding connective tissue were considerably swollen. The overall appearance was somewhat reminiscent of that of an acute allergic vascular purpura. Biopsies in control female subjects 12 hours after injection of phosphatidyl serine showed only slight perivascular cuffing.

These findings were confirmed by Agle and Wasman (1967) when in skin biopsies from patients with autoerythrocyte sensitization syndrome they found perivascular infiltration with leucocytes, and bleeding in an area of dermis deeper than would be expected after external trauma. These authors also induced ecchymotic lesions in specific locations under the influence of hypnotic suggestion in a patient with autoerythrocyte sensitization syndrome. In one experiment the patient was watched continuously for twenty-four hours to rule out the possibility that the lesion was self-induced.

From the aforementioned facts the conclusion can be drawn that the bleeding tendency in at least some cases of autoerythrocyte sensitization syndrome cannot be explained by assuming that the lesions are self-inflicted. The positive skin reactions to erythrocytes and erythrocyte stroma must be mediated by organic factors, possibly of immunological nature.

Case Reports

Two new cases of autoerythrocyte sensitization syndrome will be described, with special emphasis on the psychiatric features. Full details of somatic manifestations, laboratory findings and results of extensive skin tests will be published elsewhere (Hersle and Mobacken, 1969).

Case 1

Unmarried woman, aged 27, first treated in the psychiatric clinic for a depression in 1964. The patient and her dizygotic twin sister were adopted by a parson's family at the age of one year after her parents divorce. She has also an elder biological sister who was brought up by a paternal uncle. Her biological mother is said to have abused alcohol. The twin sister considers herself to have a nervous disposition and to be restless and irritable: she claims she has never had good contact with the foster mother whom she describes as ill-tempered and nagging. The elder sister was treated in hospital for a depression at the age of 20. She feels that the patient's tendency to touchiness, marked need for affection, sensitiveness and hypochondriacal traits are present in herself though she is better able to master her weaknesses.

The patient has always been on a very good footing with the foster father, but her relations with the foster mother have been controversial. This woman has always tended to criticize the patient who has been deeply hurt by this. Though the patient feels the foster mother misunderstands her all the time, she has always been very dependent on the foster mother; she has for example at times phoned home every evening.

The patient discontinued attending a girls' school after the fifth class (nine years of schooling). She had moderately good marks but left school because she felt one of the teachers was much too demanding. She then worked as a domestic servant and in hospitals until 1964. She had aspired to become a children's nurse but was not accepted by the training school. She worked in factories in 1966 and 1967 but was either unfit for work or in hospital for long periods during these years.

Relatives have reported that she has always been dependent on and formed attachments for elderly persons. Thus she would wish to sit on her teacher's lap when she started school. She has always demanded much attention and detailed plans to be made for her. Her endurance has been slight and she has easily given up but at the same time has been extremely thorough and pedantic. She has been oversensitive to criticism and always found it difficult to accept reprimands. From age 14 she has had difficulties with her friends whom she has tried to dominate whenever she could. With others she always feels ignorant and inferior. She has never taken any interest in boys but has had and still has girlish dreams of being waited upon by romantic suitors. Ever since childhood she has been inordinately interested in her body, felt its formations everywhere and shown up any swellings, bruises, etc. Relatives have been convinced that she has often succeeded in being admitted to hospital when for some reason she has felt lonely and isolated. Since school age she claims to have felt different from everbody else.

Medical History: Appendicectomy at age 18 (1959). Since 1960 she has been having constantly recurring very tender ecchymoses on arms and legs. From 1958 to 1968 she was hospitalized twenty-eight times, either for treatment of the recurrent ecchymoses, or in mental hospital and psychiatric clinics, or in a gynaecological department for vague acute pains in the lower abdomen. Despite comprehensive medical examinations no signs have been found of any systemic disease or haematological abnormalities. Intracutaneous injection of an erythrocyte suspension induced a characteristic ecchymosis.

The patient was first seen by a psychiatrist in 1961. She was then in a gynaecological department for abdominal pains and fever. At that time she stated she had been growing increasingly despondent and listless during the previous couple of years. She had been anxious daily, particularly forenoons and evenings. Occasionally she had attacks of feeling a lump in the throat, hyperventilation, palpitation and convulsions. She was deemed to be depressive and sent to her local mental hospital, where she

was put on levomepromazine and 'Bellergal' for ten days.

The EEG was slightly abnormal with 'moderate amplitude, a basic occipital frequency of 9 c/s with usually fairly irregular waves and a consistent pattern of slower and often triangularly rising waves with a frequency of 3-5 c/s. The latter are more pronounced on the right side and have the same or up to twice the amplitude of the background. Otherwise there are no asymmetries, no pathological complexes or notable episodes. No abnormal effects of hyperventilation'.

Thereupon, until 1964, she was in fairly good condition, when she developed a depressive insufficiency with despondency, distaste for life, lack of enterprise and increased tendency to anxiety. She also felt very irritable, lachrymose and was very hypersensitive to sensory stimuli. Thus she found it difficult to use a hair-dryer owing to hypersensitivity to heat and sound. Her memory and powers of concentration were impaired. The condition was primarily interpreted as a depressive insufficiency in a hystero-asthenic and extremely psychoinfantile, touchy and projecting person.

She was put on a course of amitriptyline and small insulin doses. During treatment marked dysphoria and tearfulness supervened. She was alternately infantile, helplessly appealing and sullenly cross. She refused to cooperate in concentration and endurance tests. She was strongly fixated on her somatic symptoms which she described in a highly dramatic fashion. She had reduced and variable muscular strength and sensitivity throughout the right arm and sometimes in the right leg.

Electroconvulsive therapy was instituted after a month in hospital and appeared to have some effects on the depressive symptoms. She found it increasingly difficult, however, to cope with institutional life. At night, for example, she wanted to get into the bed of a co-patient. She felt great need for affection but was intermittently defiant and irritable. At times she would scratch and kick, tear down the plaster and ram her head into the wall during raptus-like attacks. Owing to the suspected organic cerebral features in the symptom picture pneumoencephalography was carried out but disclosed no abnormalities.

The EEG revealed mild unspecific changes with maximum temporally and right preponderance. During sleep there were occasional hints of spike potentials but no definite paroxysmal activity.

She was discharged after two months but very soon readmitted to mental hospital and has subsequently been almost continuously in hospital for mental insufficiency or some somatic disorder despite the fact that everything possible has been done to rehabilitate her and find her a suitable job, etc. The condition seems to be more or less stationary. She finds it difficult to concentrate, for example on reading so that she now can no longer profitably read a book. In the evenings she suffers from extreme anxiety and sometimes pseudohallucinations with oneiroid features—terrifying scenes, such as funeral processions where she could identify relatives. During the spring of 1968 she was often troubled by hiccups and had daily attacks of severe dizziness, felt nauseated and vomited for no obvious reason.

She always feels inferior except on the subject of diseases and diagnostic procedures where she thinks she has superior knowledge and experience. Hence she is usually quiet and withdrawn. Sometimes, however, she makes childish and helpless attempts to win the esteem of everybody by, for example, inviting the ward personnel to coffee and cakes several days running, giving other patients packets of cigarettes, etc. She has a distinct projective tendency, usually directing her aggressiveness towards the mother and some doctors whom she thinks have 'destroyed' her.

Despite great irritability and reduced tolerance to stressful situations, she has always remarkably readily accepted various diagnostic interventions. During the past decade she has been admitted to hospital twenty-eight times but only been operated on twice. She has often injured herself by puncturing the arm veins with hypodermic needles and once demonstratively tried to burn herself with a cigarette lighter.

Psychometric Tests: IQ (SRB) = 100. Cronholm-Molander's memory test showed normal immediate memorization except for the last section of the test (human interest stories) when the patient had grown tired. Her memory after 3 hours was subnormal, the difference exceeding two standard deviations for all three sections. Benton's test for visual retention went rather poorly, the results falling in the category of 'probable organic brain lesion'. In Bourdon's test the patient made far too many omissions, indicating impaired concentration and attentiveness of approximately the same degree for meaningless and meaningful texts. Other tests, similarly, revealed little endurance, reduced concentration and memory and difficulty in managing long exercises despite great attempts to do so.

Case 2.

A divorcée of 43 who came into contact with the psychiatric clinic for the first time in 1967. The patient was born at Gothenburg where the father was a policeman. She has two older sisters, one of them with asthma. Home conditions were favourable. She was most attached to the father and since childhood had most contacts with him. The mother is described as irascible—'like me', the patient adds. She received a good thrashing once during adolescence, commenting 'I'll never forget it'. When she was about to marry the first time she could not make herself tell her mother she was pregnant but confided in her father instead.

Attended primary school for six years and a girls' school for four when she left school on account of poor marks in languages. Then she went to an arts school for two years. There she attended classes in ceramics, embroidery and sketching as well as photography. She went on to become a retoucher and copier in a photographic establishment. Has not worked for a living since 1947 but receives national assistance on account of a renal disease.

She married for the first time at age 23 and has a daughter of 19 from this marriage. The couple divorced after three years owing to infidelity of the husband. She remarried in 1951 and had three children, now aged 16, 14 and 12 years. Divorced in 1962 owing to the husband's

increasing abuse of alcohol. The children were put in her care and are well behaved. For the past year she has been living harmoniously with a widower.

Medical History: Since 1948 (age 23) the patient has suffered from recurrent pyelonephritis and from 1956 has been operated on some thirty times for renal concretions and urinary incontinence. In 1959 nephrectomy was performed on the right side owing to concretions. She was legally aborted and sterilized in 1960. Hysterectomy was performed in 1962 owing to menometrorrhagia, and in 1967 urinary incontinence necessitated cutaneous ureterostomy on the left side. On several occasions she has had acute ileus which among other things has called for explorative laparatomy. However, the latter intervention failed to reveal any cause of the pains. Since 1963 she has constantly been having recurrent haematomata on arms and legs. Intracutaneous injection of erythrocyte suspension induces a characteristic painful ecchymosis.

The patient is rather cold and rigid, principled and lacks marked syntonic personality traits. Has always been irascible and fairly irritable. This has been the cause of a few controversies among the sisters, but these rapidly pass over. Previously she was extremely thorough and pedantic, less so now. She is rather withdrawn and quiet in consorting with others. Previous case notes show that in 1958 and 1959 she had attacks of syncope and headache for unknown reasons, nocturnal breathlessness with severe oppression in the left part of the chest and severe palpitation of the heart. On some occasions she has been caught in the act of manipulating the thermometer when her temperature was being taken, and she is strongly suspected of inflicting on herself a burn or scald in the form of a red stripe on the forearm when she was about to be discharged.

In 1961 she was considered very nervous, the following year tired and somewhat depressed, in 1963 as very tired, hard and depressed. She has also been regarded as lachrymose and emotionally very unstable.

When first seen by a psychiatrist in 1967 (for suspected pathomimesis), she denied all mental disturbances of any kind apart from a slight uneasiness when crossing the street. She seemed serious, tense and expressionless but was well groomed. She was deemed normally intelligent. Her rapport with the investigator was formally correct but lacked emotional involvement and she appeared very cold and rigid and took a strongly defensive attitude. She was given an appointment at the psychiatric out-patient department but failed to turn up.

In 1968 she was felt to be hystero-schizoid, rigid and uncommunicative. Nothing but a shallow emotional contact could be established on this occasion either. Her mood was average and no psychomotor inhibition was noted. As before she denied mental disturbances even in connection with her matrimonial crises and innumerable operations, but in due course she admitted that she might have felt despondent for a day or so on being informed that yet another operation was called for. She always rapidly accepted facts. 'One is in the hands of the doctors and it's for my own sake.' She expressly denied suffering from attacks of weeping, although this is abundantly verified in previous case notes.

The EEG in 1957 and 1967 displayed a partly episodic abnormality with left preponderance with a parieto-temporal maximum and the presence of rhythmic activity with maximum in the posterior leads without noteworthy differences between sides. The EEG in 1968 was less abnormal than on previous occasions: minor unspecific deviations parietotemporally on both sides without significant side dominance.

Psychometric Tests: The IQ (SRB) was only eighty-two, viz. lower than one would have expected from her schooling. The result should presumably be interpreted as a secondary lowering of the intelligence level. Cronholm-Molander's memory test gave an average result. Other psychometric tests revealed some impairments of concentration power and memory, especially on prolonged and abstract exercises.

Discussion

Comments on diagnosis

Our two cases are in all respects characteristic of the autoerythrocyte sensitization syndrome. The sequence of events leading to characteristic painful ecchymoses, the associated symptoms in the form of abdominal pains and menometrorrhagia, the response to intracutaneous injections of erythrocyte suspension, and the non-contributory haematological findings, fully accord with the original description by Gardner and Diamond (1955).

Initially malingering was highly suspected, but close observation at several hospitalization periods did not indicate that the lesions could be self-inflicted. Intradermal injections of autologous and homologous erythrocyte suspension always resulted in painful ecchymoses if the concentration was 20 per cent or above. We could exclude self-induced artifacts by watching the patients continuously. Control testing was performed with normal saline, serum, plasma, autologous white cell and thrombocyte suspension, histamine and calf thymus DNA. All reactions were negative. Furthermore, ten control patients were tested with erythrocyte suspensions of different concentrations with negative results.

To elucidate further the specific reactivity in the two patients serial skin biopsies were taken after intradermal injection of erythrocyte suspension. Already after three hours an intense vasculitis with infiltration of neutrophils and eosinophils had developed.

A characteristic feature was the prolonged course. Treatment has so far been of no avail in

our cases and after eight and five years, respectively, the patients still have their ecchymoses.

Psychiatric comments

Case 1 exhibits marked hysteroid personality traits (Lindberg, 1963) with fickleness, impulsiveness, egocentricity, tendencies to anxiety, autonomic instability and hypochondria. She possesses the hysteroid person's shaft vision and constantly demands attention. She is also markedly psychoinfantile. Since childhood she has been helpless and dependent. Her sensitivity is considerable. She has always felt inadequate and inferior and feels that her mother has aggravated this by incessant criticism. Moreover she has had an astheno-emotional syndrome for many years with marked concentration impairment, memorization difficulties, great irritability, and from time to time affective explosiveness. She tires easily, is lachrymose and hypersensitive to sounds. These symptoms may be interpreted as features of an organic cerebral pattern (Bonhoeffer, 1912). Her hysteriform symptoms lack definite organic features, but, on the other hand, some form of cerebral lesion cannot be ruled out. It is well known that so-called hysterical symptoms may be associated with various cerebral lesions. Follow-up examinations of some series have revealed that such lesions have been present in the majority of consecutive series of patients with hysteriform symptoms (Slater and Glithero, 1965; Whitlock, 1967).

The most striking feature of Case 2 is her vehement denial of having any symptoms of mental insufficiency, although such manifestations are well documented in her history. It seems likely that her denials are the result of a hysteroid repression. One gets the impression that she has hysteroid personality traits, but a schizoid rigidity and sensitivity is undoubtedly the dominating factor. She strongly defends her integrity, is unwilling or unable to admit her weaknesses. The patient has shown some astheno-emotional symptoms, including tiredness, irritability and tearfulness. The intelligence test produced a remarkably poor result compared to what one might have expected, which suggests a secondary lowering of the intelligence level. The abnormal EEG is another indication of cerebral dysfunction of unclear nature. Interestingly, Case 1 also had an abnormal EEG. The EEG has previously been recorded in only two cases of autoerythrocyte sensitization syndrome (Reed and Firkin, 1957; Waldorf and Lipkin, 1968), and in these cases too there were EEG abnormalities.

Our Case 2 has a peculiar surgical history, with some thirty operations, most of them on the urogenital tract. She has, too, a strong attitude of 'grin and carry on' and in conversation belittles the strains the many operations must have caused while tending to pose as something of a martyr. Similar reactions have been reported by Agle and Ratnoff (1962). In fact, both cases have in common an adaptation to hospital life. The frequent operations and periods of hospitalization seem to have been justified when considered separately, but the overall impression of the histories is unnatural, a point where both cases agree well with published data. Notably it has been demonstrated that surgical overtreatment is positively correlated with a hysteroid personality (Lindberg and Lindegård, 1963). This is probably because hysteroids intensely experience somatic troubles and all kinds of pain and are entirely absorbed by them. The 'hysteroid lens' magnifies the sensation of pain and trouble, to all of which must be added the low anxiety threshold.

The patients' emotional peculiarities have made it hard for them to maintain normal interpersonal relationships, especially towards relatives. There are some striking similarities between the patients. Case 1 is unmarried and Case 2 divorced twice. They were both the youngest in their families and both of them clearly showed a preferential attachment to the father or his substitute. Over the years, emotional stresses of various kinds have been very common occurrences. However, no conclusive parallellism has been detected between increased haemorrhaging tendencies and particularly strong emotional stresses. Notably during recent months, however—when we have had close contact with her—Case 2 on two occasions had particularly large and tender ecchymoses, both times contemporaneously with emotional stress (once when she watched over her seriously ill mother, the other time when

her spouse temporarily left her). In all essentials our two patients exhibited symptom pictures and personality patterns that agree well with the peculiarities reported in previous cases. Despite different frames of reference and terminological dissimilarities it is evident that the phenomenological characteristics are strikingly similar in the thirty-three cases of autoerythrocyte sensitization syndrome reported so far.

The association between autoerythrocyte sensitization syndrome and particular psychopathological features

It seems well established that autoerythrocyte sensitization syndrome is very closely associated with a particular type of personality and particular psychopathological phenomena. In various diseases with concomitant cerebral lesions an equally strong correlation may exist between particular somatic signs and mental symptoms. This is so in, for example, general paralysis, such severe endocrine disorders as hypothyroidism and hyperthyroidism, Addison's disease, and a variety of genetically determined oligophrenic syndromes. In some other illnesses unaccompanied by cerebral lesions there may also be a statistically significant association between a given personality type and a given disease. Duodenal ulceration is a well-known example of this (Wretmark, 1953), but these constitutionally dependent relationships are characterized by the fact that they are much weaker than the surprisingly strong association between autoerythrocyte sensitization syndrome and particular personality traits. Accordingly one must consider in the first place whether it could be a matter of an artificial association, something which might be brought about in a number of ways.

1. What first comes to mind is that the lesions could be self-induced. A high proportion of these hospital-addicted, help-seeking, hysteroid persons call to mind the Münchhausen syndrome. This problem has already been discussed in the literature review and in the comments on somatic diagnosis above. It must be considered proved that self-induced lesions are most unlikely in the causation of autoerythrocyte sensitization syndrome.

2. Self-inflicted haemorrhages might be of secondary significance through accelerating any immunological-vascular abnormalities such patients might have. If so it would be reasonable to expect hysteroid psychoinfantile patients who have difficulties in coping with the stresses of life to be over-represented among the more severe cases of the syndrome.

3. It is conceivable that the thirty-three cases reported up till now over a period of twelve years are not representative. Autoerythrocyte sensitization syndrome is very probably more common than this figure suggests; and if so it is not unlikely that extremely helpless patients with hysteroidly magnified sensations of their experiences would be over-represented among the first thirty-three cases diagnosed.

If these artifactive explanations can be ruled out—that is, if the cases described so far really are representative of a nosological entity in the usual sense of the term—the association may be accounted for in various other ways. One possibility is that a cerebral lesion of some kind is the connecting link. This is suggested by the presence of EEG abnormalities and an astheno-emotional component in the clinical picture as well as by the occurrence of hysteriform manifestations. As pointed out by many workers (Kennedy, 1940; Slater and Glithero, 1965; Whitlock, 1967), so-called hysterical symptoms are often associated with cerebral lesions. Cerebral haemorrhages could conceivably occur in auroerythrocyte sensitization syndrome, just as there are haemorrhages in the skin and in the urinary and gastrointestinal tracts.

Nevertheless it seems as though the personality traits and the psychopathological manifestations associated with autoerythrocyte sensitization syndrome develop long before manifest haemorrhages. Hence one may speculate whether in these cases there exists some form of genetically determined cerebral dysfunction giving rise to a particular type of mental vulnerability and deviant personality development. Such a genetic background might also lead to other disturbances through pleiotropic effects, for example defective coagulation, perhaps by way of immune reactions. This would account for the very strong correlation between autoerythrocyte sensitization syndrome and a particular psychiatric picture.

Needless to say this hypothesis in no way excludes other mechanisms, e.g. physical trauma, emotional stress, or specific conditioned mechanisms as the precipitating factor. Yet the hypothesis does provide a possible model for explaining why a few persons with the personality structure described develop autoerythrocyte sensitization syndrome.

Summary

Two new cases of autoerythrocyte sensitization syndrome are reported, with special emphasis on psychiatric aspects. The relevant literature is reviewed. The patients exhibited hysteroid, sensitive and psychoinfantile personality traits, inadequate social adaptation and mental insufficiency with, among other things, hysteriform manifestations and autonomic dysfunction. It is emphasized that cerebral damage may have been present. Explanatory models are presented to account for the observed association between the specific somatic symptom picture and the fairly consistent psychiatric features.

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