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REMARKS ON THE VARIETIES OF CHRONIC CHOREA, AND A REPORT UPON TWO FAMILIES OF THE HEREDITARY FORM, WITH ONE AUTOPSY.[1]

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NOTHING illustrates so pointedly the widespread interest now taken in diseases of the nervous system than the rapid manner in which facts accumulate about obscure and rare affections. Twenty years have passed since Huntingdon, in a postscript to an every-day sort of article on chorea minor, sketched most graphically, in three or four paragraphs, the characters of a chronic and hereditary form which he, his father and grandfather had observed in Long Island. In the whole range of descriptive nosology there is not, to my knowledge, an instance in which a disease has been so accurately and fully delineated in so few words. No details were given; the original cases were not even (nor have they been) described,[2] but to Huntingdon's

[1] Read before the Philadelphia Neurological Society, Nov. 28, 1892.

[2] Several years ago I made an attempt to get information about the original family which the Huntingdons described, but their physician stated that, owing to extreme sensitiveness on the subject, the patients could not be seen.

account of the symptomatology no essential fact has been added. Within the past eight years a copious literature has gathered around the subject (particularly in this country), which is available to 1889 in the monograph of Huet.[3] Since this date the interest has even increased, and the references stand thick and close in the Index Medicus for the past three years. The recent paper by Sinkler (*Medical Record*, March 12, 1892) gives the literature to date. The practical outcome is that we now know the clinical aspects of this form thoroughly, and I have nothing unusual to offer in the history of two Maryland families which I have to report; but the connection of the chronic choreas with each other and their relation to chorea minor are questions which may be discussed, and upon which we need fuller information.

A chronic chorea of adults and aged was recognized long before Huntingdon's description of an hereditary form in adults, which was itself antedated in this country by the observations of Waters, Gorman, and Lyon.

Provisionally, at least, we may place the cases of chronic chorea in four groups:

First group, chorea of infants, appearing either at birth or within the first two or three years of life. Until recently but little attention has been paid to these interesting cases, of which there have been several well-marked examples at the Philadelphia Infirmary for Nervous Diseases; one has been reported by Sinkler and two by myself. A *résumé* of the literature to date is given by Audry in his recent monograph upon "Double Athetosis." The cases heretofore described may be, as he says, divided into those in which no accurate account is given as to the existence of spasm with the movements, and those with explicit statements as to its presence or absence. A majority of these cases are examples really of spastic diplegia, plus movements which may be choreiform, tremulous, or athetoid; or there may even

[3] De la chorée chronique, Paris, 1889.

be combinations of mobile spasm with more rapid movements, so that the diagnosis is extremely difficult, one observer calling the case chronic chorea, another double athetosis. This confusion was well illustrated in the discussion at the Berliner Gesellschaft f. Psychiatrie u. Nervenkrankheiten last year, when Remak showed a case of chronic chorea which Oppenheim had regarded as possibly athetosis, and which Senator thought—owing to the existence of spasm—had nothing whatever to do with chorea. In a large proportion of these cases there is also mental impairment, or even idiocy. The following case illustrates choreiform movements in a child with extremely slight spastic manifestations.

Female, aged four and a half years, seen in Ontario, May 12, 1892. One of twins, born prematurely at the eighth month. Mother had one child before, also at eighth month. Nothing abnormal was noticed about the baby at birth, it was not blue, and subsequently throve well. No abnormality was observed until the other child began to creep, when this one seemed backward and could not hold on to anything. At one year irregular movements were noticed in the arms and legs, and have continued. Teeth were cut at the twelfth month, and she began to talk at the third year; has never walked. The child is bright, intelligent-looking, with well-formed head; does not dribble. There is no nystagmus; talks a gibberish, of which I can only catch a word or two, but which the mother understands quite well. Movements slight in face, scarcely noticeable; no distinct grimaces; movements of tongue natural. The arms are in constant motion, slow and irregular as a rule, but occasionally jerky in trying to grasp objects. The fingers do not display athetoid movements. She cannot use a spoon, but can feed herself with bread, etc. The mother is sure that the arms are never stiff. She sits up well, but the head occasionally comes forward with a jerk. The feet are extended in talipes equinovarus position, and the toes spread occasionally in athetoid movements. The legs are freely movable, not apparently stiff; the muscles hard, but not very well developed. In taking off the stockings, however, the legs stiffened and were hard to bend at the knees, and the big toes became strongly flexed.

This case, belonging to the group described in literature as chorea spastica, is more properly a spastic pāraplegia with choreiform and athetoid movements. The following is an illustration of a less common type, in which there was no spasm and the movements were of a more characteristic kind.

N. G., aged eight and a half years, the eldest of two children. The mother had twitching of the eyelids when young, but there are no nervous troubles of any moment in either her own or in her husband's families. The patient was a delicate infant, but throve fairly well, learned to walk and to talk at the usual time. About the fourth year it was noticed that she had irregular jerking movements in the arms, which were moved about wildly and even thrown over the head. She became excitable and irritable, and slept badly. Within a few months the face became affected, and she made grimaces, and sometimes a peculiar grunting noise. The legs were involved shortly after the face, and at times she walked with difficulty. When seen in 1890, more than four years after the onset, she seemed a well-grown child for her age, was not anæmic, a little nervous in her manner and excitable, but intelligent looking. After sitting quietly for a few moments, the arms jerked about and the face twitched. The right arm is most affected, and is twisted about in an odd way, and lifted as high as the shoulder. The legs are now not much, if at all, affected, though she fidgets about in her chair. When watched, the movements are much increased. She feeds herself with great difficulty. There is no spasm in the muscles, which are well nourished; the reflexes are not increased. There is no heart affection. Treatment has not been of the slightest benefit. She is very wayward; and though bright mentally, it is difficult to get her to attend to her studies. There have been no explosive utterances, or any of the mental features of convulsive tic.

And lastly, some of the cases of chronic progressive chorea with dementia have begun in early childhood.

Second group, comprising cases of chronic chorea without any hereditary *anlage*, in which the disease may set in in childhood, adolescence, maturity, or old age. Many of the cases in Huet's monograph had no history

of chorea in the ascendants. In scarcely any of the features are these cases to be distinguished from the variety described by Huntingdon, but in many instances the disease has begun in childhood or adolescence, and has gradually led, in a variable period of time, to dementia. Very many cases of this kind have been reported recently from asylums.

Only some of the cases of chorea in the aged can be classed here, since many run an acute course, and recovery is not uncommon, noted indeed in eleven instances in Herringham's critical review upon chronic chorea, in *Brain* (1888). The acute course, and the association occasionally with rheumatism, render it probable that many of these are really instances of chorea minor.

Third group, including the cases with marked heredity, the so-called Huntingdon's chorea, characterized by the occurrence in family groups, a late onset, psychical disturbances, and a progressive and fatal course.

Fourth group, comprising cases of chorea minor which pursue a chronic course, and persist for months or even years, and ultimately recover. They differ essentially from the other forms we have been considering, in the absence of a progressive character, the more active, quick, bizarre movements, and the retention of the mental powers. The following is a good illustration of the chronic form of chorea minor:

Alfonzo G., aged twenty-one, baker, applied to the Infirmary of Nervous Diseases, June 1, 1885, with spasmodic movements of the muscles of the face, arms, and trunk. The affection had lasted without intermission since August, 1884. There was no rheumatic history in the family, but a sister had chorea, and subsequently died of heart disease. He is a strongly built, well-nourished young man. The muscles of the head and neck and those of the face contract suddenly, jerking the head upward and rotating it slightly. At the same time he makes a grimace, and the muscles of the thorax are thrown into quick action, and the air is drawn in often with a whistling sound. The heart is not involved.

The patient was under observation and treatment for three years, during which time the chorea persisted with

slight variations in the intensity of the movements. When I last saw him the twitching and jerking of the muscles of the neck and chest were present, but the facial spasm had lessened. There were no mental symptoms, and for the greater portion of the period he was able to work.

Other instances of chronic chorea minor in the records of the Infirmary are given in my "Lectures on Chorea,"[4] two of which are very interesting from the persistence of the symptoms for more than three years with ultimate recovery.

Habit spasm, beginning in childhood, may persist for years, and is often confounded with chorea minor; there are also aggravated forms of convulsive tic with movements typically choreic, but which can usually be separated from chorea minor by the existence of fixed ideas, coprolalia, etc.

The following is a record of two family groups of the hereditary form of chronic chorea. The cases present the usual peculiarities described by Huntingdon. For the opportunity of seeing the members of the first family and for the details of the pedigree, I am indebted to Dr. Ellis:

FAMILY X.—FIRST GENERATION.

A. B., an Englishman, married C. D., a native of —— County, State of ——, and had of issue eleven children. A. B. died aged eighty-seven, and his wife aged eighty-five. Neither of them, so far as is known, displayed any mental or bodily peculiarities. Two of the eleven children died choreic and demented.

Of the other children, two of the girls married N.'s. One died aged seventy-five, leaving children, all of whom are in good health; the other, Mrs. N., still lives, aged seventy-seven, and has healthy children: George, died aged seventy, a bachelor; Sarah, died aged fifty, of typhoid fever, without issue; William, died aged seventy-six, leaving a large family, none of whom have shown any symptoms of the disease; Mary, died of an acute illness, aged fifty-five, leaving healthy issue; Jane, died

[4] Medical News, Philadelphia, October, 1887.

aged seventy, leaving a family, none of whom are affected; two other daughters died maidens, well advanced in life. The two affected children were James and Margaret.

James, the first to become affected, began to exhibit remarkable muscular irregularities before he was forty. Dr. Ellis writes: "I very well remember, in my earliest youth, his grotesque movements, exciting unusual attention, and I fear more ridicule than sympathy. His swaying, jerking, and fantastically irregular walk compelled him from the sidewalk to the unobstructed roadway. Notwithstanding his infirmity, he was a great pedestrian, frequently walking from his home, eight miles distant, and returning the same day. His sudden stops and precipitate advance, his facial contortions and mobile features, I recall with great vividness after forty years. His wife died in childbed.

Margaret, married J. M. Her symptoms began to develop before she was forty. She continued to go about until a few days before her death, which occurred in her sixty-fifth year. Except a short time before her death, she was not entirely helpless, nor were the mental symptoms very strongly marked in her case.

SECOND GENERATION.

Margaret M., the last-mentioned patient, had five children, two of whom have already died of the disease, and three are in various stages of it. I have seen two members of the family, and have performed a post-mortem on a third:

First child, male, now in his sixty-first year. A year ago the first evidences began. "A man of some character, it is but charity to ascribe the eccentricities of his life to disturbed mentality. He married twice, but had issue only by his first wife. Several children died in infancy, but one surviving is now in good health." This patient I could not see.

Second child, female, married, became choreic in her fortieth year, and died demented in December, 1890, in her fifty-eighth year. She was confined to her bed for nearly a year before her death, which occurred in the Pennsylvania Hospital for the Insane, Norristown. She had four children—three girls and one boy; all are living and in good health, the oldest being now in her thirty-second year.

Third child, male, aged fifty-five. I saw this patient with the doctor. He has enjoyed good health, and has been able to attend to his business until recently. When about forty-two he began to get nervous. Irregular locomotion was the first symptom; his speech became affected about a year ago. He will make use of a nod or a grunt in place of words whenever he can. Lately he has been confined to the house, and has been obliged to abandon business. He is very irritable, and is steadily passing into a state of dementia. He has had five children: four are living and in good health, the oldest about thirty-three years of age; one died of basilar meningitis at sixteen. I saw this patient in April, 1889, and made the following note:

Bony, well-built man; face has an intelligent expression. The gait is very peculiar; he sways from side to side; the movements are irregular, very unlike those of an ataxic, but resemble rather those of an alcoholic. He does not use a cane; feet are not specially spread; eyes not directed to the ground. He can stand with his heels together, with his eyes shut; no movements of the hands or arms when at rest, but in attempting to move there are large irregular sweeps of the arms and slight tremor. He has great difficulty in feeding himself, and sometimes takes two hours or more at a meal. He still can write, though with increasing difficulty. He signs his name to a letter, but the pen, in forming the letters, is often jerked up and the signature is very irregular. With the eyes shut he touches the nose or ear with precision and quickly. The grasp of the hand is firm and strong. There is no disturbance of sensation, no numbness or tingling. Knee-jerk slightly increased; ankle clonus not obtainable. Pupils medium size; react to light and on accommodation. Speech is slow, and interrupted frequently by the interjection 'Hem, ha!' This peculiarity, his wife says, is of comparatively recent development. The mental condition is apparently good; perception clear. When questioned, however, on several occasions, it seemed to take him some time to understand our wishes. He takes an interest in what is going on; reads a good deal, particularly the newspapers. He still personally conducts his business.

Within the three years and a half since making the preceding note he has steadily declined mentally and bodily.

Fourth child, female, aged forty-three, married, has had five chidren. One died of scarlet fever; the others

are living, the oldest a boy of twenty-three. In this case the disease has progressed with greater rapidity than in the others, and certain indications of it have been present, according to the doctor, since her thirty-fourth year. The mental symptoms were first to appear. In April, 1889, I made the following note:

Slightly built, somewhat anæmic woman; talks clearly and rapidly, but occasionally she displays a certain childishness, and the doctor, who has not seen her for some years, was much struck with the change in this respect.

While sitting quietly there were no irregular movements of her limbs, but occasionally there was a slight jerk of the finger, the shoulders would move, and once or twice, while speaking, there appeared to be irregular contraction of the facial muscles. There is no tremor of the tongue, and the pharyngeal muscles act normally; the grasp is good; she can use her fingers for delicate movements, and can thread a needle, and there does not appear to be the slightest inco-ordination. The most marked change appeared to be noticed in her gait. She walks with the feet somewhat spread, but follows a straight line fairly well; she turns with a little difficulty, and, if rapidly, loses her balance. Her head is carried somewhat stiffly in walking; she does not trip, and she walks in the dark quite well. She stands with her eyes shut and her feet together without swaying.

The power of the legs is good; knee-jerk increased on both sides; no disturbances of sensation; special senses normal; the pupils are of medium size and react to light and on accommodation. In the three years since the preceding note was made she has lost ground rapidly, and the muscular inco-ordination has become much worse. She is now confined to the house, and for the greater part of the time to her bed.

Fifth child, female, aged, at the time of her death, fifty-one; married; had eight children. Dr. Ellis writes: "After the birth of seventh child, in her thirty-second year, her husband noticed the beginning of the trouble in jerking movements of the legs when sitting, and when erect she had a trick of raising her heels suddenly and standing upon the ball of the toes. Irregular movements of the arms speedily followed. When I saw her first, in 1880, she could walk a mile or two without apparent fatigue, and would insist on walking to church, nearly a mile distant, repelling the suggestion that she could not walk as well as another. At this time, in walking, her

body would be bent forward, her head jerking, with a pendulum-like motion, to and fro, and her legs making such irregular and large movements that she would make wide excursions on the sidewalk. A year later she could no longer go out without assistance. Her speech indicated marked changes very early, in her fortieth year, and this was (in 1881 and 1882) accompanied by great difficulty in swallowing and frequently with alarming spells of strangling. She was a most pitiable sight. She suffered also from procidentia uteri; yet in June, 1883, in her forty-third year, she was delivered of her eighth child, which survived but a few days. Her menses were perfectly regular, her menopause occurring in her forty-eighth year. Six months before her death she was confined to her bed, utterly helpless, and was fed with a spoon. She was now entirely demented.

"Her deep reflexes were rather exaggerated. She could go about the house at night with as little help as in the daylight. She was exceedingly irritable and cross. The choreic movements stopped in sleep; there was no palsy of the sphincters. Of her eight children, seven are living, the oldest in her thirty-third year; all are in good health."

Post-Mortem (about thirty hours after death).—Considerable wasting of the body; no enlargements of joints; no abnormal position of limbs; face a great deal wasted, presenting several recent scars and abrasions, the result of falls.

The skull-cap of moderate thickness; dura tense; meningeal vessels looked stiff; longitudinal sinus contained recent clots. On the exposed cortex cerebri the arachnoid was somewhat turbid and universally separated from the pia by a considerable layer of serous exudate; this was especially marked over the sulci. Pacchionian granulations were numerous; cortical veins moderately full. At the base the arachnoid was turbid and the larger arteries a little stiff; the meninges were not especially adherent, and the pia could be stripped without tearing the substance. Superficial examination revealed no areas of softening, and no special lesions of hemispheres or of cerebellum. There was general wasting of the convolutions, which were also, on section, rather firm. The gray matter was dark, and in places looked thinner than normal. The crura presented no signs of descending degeneration; the pons and medulla were natural-looking; anterior pyramids had a clear, normal

aspect; the ventricles were not distended. Spinal cord was firm; arachnoid a little opaque; pia normal. Transverse sections showed no systemic degenerations; the gray matter had a rosy red tint.

Microscopical Examination.—I am indebted to Dr. Gray for an extensive series of sections from various parts of the brain and cord. The changes may thus be summarized: The arteries were thickened and in places showed hyaline degenerations, and, in the smaller arterioles, fatty changes, very marked in the fresh specimens from the cortex. Here and there the perivascular lymph-spaces were large and contained leucocytes. The ganglion cells in many sections showed very slight changes, not more than are often seen in chronic disorders associated with atrophy of the convolutions. There was the common vacuolation, and many cells seemed laden with pigment. The increase in the connective-tissue elements was more evident to the touch and on section than microscopically. Sections of the pons and medulla showed no special foci of disease. Beyond thickening of the arteries and a shrinkage in the cells of the anterior cornua (probably an artificial change), the sections of the cord showed no important lesions.

The morbid anatomy of chronic chorea is that of a neuro-degenerative disorder—diffuse changes in vascular, ganglionic, and neurogliar tissues—not essentially different to, though less pronounced than, those of dementia paralytica. We see, too, the terminal series of the process, far removed in time from, not necessarily akin to, the initial alteration which lies at the basis of the disordered function.

The doctor writes that, prior to the onset of the chorea, "these patients and their children are intelligent and bright, and the women are comely. The men are rather aggressive, energetic, and ferocious; the women are affectionate and prolific: the issue of the five numbers twenty-seven. There is no history of infantile chorea in the family, nor of rheumatism, nor of heart disease. The period of development of the symptoms covers a wide range, from the twenty-second to the sixtieth year. The symptoms have begun earlier in the women than in the men. There is at present no sign of disease in any

member of the third generation, though several of the children are past thirty-five. There seems to be a remarkable insensibility to pain in these cases; they fall about and bruise themselves severely without complaint. Shortly before the death of No. 4, she struck a cast-iron key, lodged in the door-lock, with her hand and broke it, naturally bruising and maiming her hand very much; but of this she took no notice whatever. The uncle and the mother of these patients kept about and showed much greater muscular vigor than members of the second generation, in whom, too, the dementia has apparently progressed more rapidly. The progress of the disease is marked by great emaciation; the movements are but little under control of the will and are much excited by volition. When standing, only those muscles are much affected which are concerned in balancing the spinal column and the head; the movements stop during sleep. These patients have all been light sleepers. The speech defect is not aphasic, but muscular —an indisposition to articulate on account of difficulty in moving the muscles. In case No. 4 the symptoms were very similar to those of a case of bulbar paralysis."

NEITER FAMILY.

So far as can be ascertained only four members of the family have been affected, namely: mother and three children, one of whom was our patient, Peter.

1. The mother, a German, is stated to have had trouble of the same kind as that which Peter has. For many years she made wild inco-ordinate movements with her arms, and toward the end of her life she could not eat alone and had to be fed. Her mind, also, became very weak. The exact duration of the disease in her case could not be obtained, but it extended over several years. She is said to have died of heart disease. She has one brother living, aged eighty-three, who is said to have the disease, but Dr. Simon visited him and reports that he is only subject to ordinary senile tremor. No information is available with reference to her family. Her maiden

name was Schmidt. She had four children, of whom three have been affected with the disease.

2. Lizzie N., was well up in her thirty-seventh year; married and had six children, of whom two died and four are living and well. After the birth of her last child the chorea developed, beginning in her arms first. Her husband noticed that she frequently dropped things. The trouble gradually became worse. Her mind became seriously affected, she talked incoherently, and had strange ideas. She once tried to commit suicide by jumping out of a window. The last year of her life she was helpless and could not walk alone. She died in her forty-ninth year, about twelve years after the first onset of the symptoms. Her husband, from whom these facts were obtained, says that the disease looked very much like St. Vitus' dance.

3. Nicholas Neiter, aged about forty, blacksmith, living at Edgewood, Hartford Co., Md. He was seen for me by Dr. Chas. Simon, who reports that he is evidently subject to the disease, as he displays grotesque inco-ordinate movements of the legs, arms, and face. Mentally, too, he is inclined to be childish and is very emotional. He regards himself, however, as in a condition of perfect health and not affected in any way as his brother Peter.

4. Peter Neiter, aged fifty-nine, German, a butcher, was admitted to Johns Hopkins Hospital,[5] October 9, 1890. Patient has been in this country since 1850. He has always enjoyed good health with the exception of malaria when he first came to this country; has not had syphilis. He dates his present trouble from an attack of gastro-intestinal disturbance eight years ago, which followed the drinking of large quantities of iced lemonade. At this time he had also pains in the head, and he speaks of the occurrence of something bursting in his body like a cannon. The movements began about five days after this over-heating and taking iced drinks. They did not start at any particular part of the body, but were general from the outset. They have gradually become worse, particularly when voluntary movements are made. They are severe enough to prevent him from working, and he has not been able to do much for six or eight years. He has fallen, sometimes, owing to the irregular movements of the legs. He has never at any time lost consciousness.

[5] The patient was shown at the Hospital Medical Society, and is reported in the Bulletin, vol. i.

Emotion or fright always exaggerates the movements. He has not had headaches; has as a rule slept well. His appetite has been good and general health excellent. Ever since the attack, eight years ago, he has been liable to a recurrence of the vomiting whenever he takes cold drinks. He says his memory is quite good. He does not think that his speech has been affected.

Present Condition.—The patient is a large, well-nourished, well-built man. The face in repose looks intelligent, but on smiling, the expression is fatuous. He answers all questions readily and freely; gives a good account of his condition, and it is more in his expression and general behaviour that an indication is found of mental impairment.

When sitting in a chair, at ease, the arms and hands are in more or less constant irregular motion. The fingers are extended and flexed alternately; sometimes only one, sometimes the entire set. At other times the whole hand will be lifted or there are constant movements of pronation or of supination. For half a minute or so they may be perfectly motionless. The head and trunk present occasional slow movements; in the latter more of a swaying character. The legs jerk irregularly and the feet are flexed or extended; but the movements are not so frequent as in the arms. The face in repose is usually motionless, but the lips are occasionally brought together more tightly and the chin elevated or depressed. There is an occasional movement of the zygomatic and of the frontal muscles. He puts out the tongue, with tolerably active associated movements of the face, and it is usually quickly withdrawn or rolled from side to side. It is impossible for him to hold it out for any length of time. There are no irregular movements of the palate muscles.

He walks with a curious irregular gait, displaying distinct inco-ordination, swaying as he goes, hesitating a moment in a step, keeping the arms out from the body and in constant motion. The legs are spread wide apart; steps are unequal in length and he seems rather to drag the feet. He stands well with the heels close together.

There is a suggestion of stiffness about the gait and about the way in which he uses his legs.

Sensation is unaffected. The deep reflexes are increased. There is slight ankle clonus, exaggerated knee-jerk, and slight increase in arm-reflexes.

The special senses are unimpaired. Pupils are of

medium size—the right a little larger than the left; they react to light and on accommodation; there is no nystagmus. He has no fever; bowels are regular, and the urine shows no special changes.

A report of cases of the hereditary form of chorea does not afford a very wide scope for discussion; but there are problems in the relation of the forms to each other and to chorea minor, which, if I have read the literature aright, are still far from settled. My own point of view may be very briefly stated: Chronic progressive chorea is a malady distinct from the various disorders associated with coarse lesions of the motor centres or path known as symptomatic chorea—an affection which (like forms of muscular atrophy) may occur in families or in single individuals, and is characterized by irregular, inco-ordinate movements, a reeling gait, speech disturbances, and progressive impairment of the mental faculties. The movements differ from those seen in chorea minor, being slower, and resembling rather those of Friedreich's ataxia, without the brusque, jerky character of the former disease. Moreover, in striking contrast to the movements of chorea minor, those of chronic progressive chorea are sometimes influenced by the will. A certain number of the cases of chronic chorea beginning in infancy and childhood belong to this category, but a very much larger number are instances of spastic paraplegia or diplegia; while others represent anomalous forms of chorea minor.

Chronic progressive chorea is, I believe, a disease wholly apart from the affection described by Sydenham, having nothing in common with it but the name. The course of acute chorea minor, the incidence in children, the arthritis, the seasonal relations, the extraordinary frequency of endocarditis—to say nothing of the different characters of the movements above referred to—separate it as a well-defined affection, depending possibly on a virus as yet unknown.