CHOREA, TICS AND COMPULSIVE UTTERANCES.*

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ACCORDING to the accepted description, a tic is a psychogenic symptom, an expressive movement derived from a mental fixation. This concept was rejected by some of those authors who observed tic-like movements as sequelæ of acute encephalitis. Other cases, quoted by Steck (1), in which the onset of a hyperkinetic encephalitis was associated with tics, suggested still more definitely that there might be an organic cause for such movements. An extreme concept localized the lesion responsible for these symptoms in the corpus striatum. This wholesale assumption was in turn objected to from the neurological standpoint, particularly by Kinnier Wilson (2), who, in his well-known paper, pointed out how complicated the possible connections were. In particular he mentioned that even tics following encephalitis might be conditioned not only by the brain disease itself, but also by constitutional and psychic factors. The network of different ætiological factors becomes less complicated when a disease like chorea is studied, rather than cases of encephalitis where widespread permanent lesions remain. We have, therefore, studied the occurrence of tics in connection with chorea, though we do not claim that any conclusions can be drawn from these observations for tic neuroses generally.

It is now generally held that tics, even the *maladie des tics* of Gilles de la Tourette, do not constitute a disease entity. In 1902 Meige and Feindel (3), writing on tics, noted that torticollis may occur after chorea. Oppenheim (4) made similar observations, although neither drew any general conclusions from these facts. In 1927 one of us (5) published histories of cases of Sydenham's chorea showing a surprising frequency of motor after-effects. Since then three other authors have discussed the same question. E. Straus (6), Ossipowa (7) and Kraus (8) have submitted material corroborating that, in a high percentage of chorea cases, motor disturbances, frequently in the nature of tics, remain. From the opposite approach E. Straus also collected seventeen cases of tic, and found in the early history three instances of chorea, and five

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others following tonsillar infections. On going through the histories of in-patients at the Maudsley Hospital, we found during the years 1932 to 1935 fourteen cases in which tic was noted in the diagnosis, and of these six had a history of previous chorea. (This is a selection, in that many other instances of tic were probably observed as a minor symptom, but were not referred to in the final diagnosis.) The connection is too frequent to escape attention, and renders difficult the conception that the origin of tics can be a purely psychogenic one. On the other hand, there are many arguments against the assumption of a focal recurrence of chorea, in particular the appearance and disappearance of the symptom under emotional influences, or its coming and going in association with a general disturbance, such as a depression. Among these fourteen cases, in more than one a recurrence of tics was observed during the course of a depressive illness.

The connection between tic and chorea does not decrease the importance of constitutional factors, even though chorea may be an infective disease. Since our previous investigation the hereditary predisposition to chorea has been supported both by systematic studies of heredity (B. Schulz (9)) and by numerous published familial cases.

The existence of a predisposition to tic has frequently been claimed by authors, but has never been exactly defined. Cases with familial occurrence are too suspect of being produced by imitation. Our observations raised the question whether we see this predisposition in the constitutional anomaly claimed for chorea, or in the post-choreic motility disturbances. We are more in favour of the latter, since we have often observed tic following chorea, but never chorea following tic. Otherwise expressed, we regard the predisposition towards tic in our cases as an acquired one, due to the chorea. Nevertheless, the other possibility, namely a common hereditary constitution, is sometimes suggested ; there is among our cases of tic without chorea one in whose family chorea was present.

Nevertheless we hold that the neurosis chooses the organ (using that term in the Freudian sense) on account of the motor lability, acquired or given by constitution. The question also arises whether the determination of the particular tic depends on other than psychic factors—in other words, whether the tic always has an expressive value (or has had it at any stage in its development). For the discussion of this question, tics with verbal utterances are conspicuously interesting, as in these the expressive character is especially clear.

Straus (10) has discussed physical and psychic aspects in a case of tic with coprolalic utterances following on a chronic chorea. He was able to demonstrate that in this case articulatory and respiratory hyperkinesias were the pathological basis of the coprolalia, and that they were only secondarily modified by psychic elements.

At the Maudsley Hospital six cases of phonatory respiratory tics following

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chorea were collected, which to some extent illustrate the separate stages of this development. In five cases we obtained definite histories of one or more attacks of chorea. The sixth, a boy, æt. 10, became ill with a choreiform motility disturbance after a shock sustained when he was five. He and another boy who were examined in detail, showed irregular partial contractions of the diaphragm, twitchings of the thoracic muscles, and myoclonic movements of the abdominal muscles which we were able to film. They resemble in every way the pictures taken by Reisch (11) in his case of post-vaccinal encephalitis. This similarity suggests that this sixth case also has an organic origin.

All our cases produced grunting and " barking " noises, and in two cases these were the only noises produced. One had recently began to mutter and to repeat remarks under his breath, this only happening at home. In another case (A. S—) we heard only words uttered compulsively with the tic in a low voice which was barely audible. They had no intelligible sense, but at home, and at school, where the boy was teased, irrelevant obscenities were overheard. In another case (M. S—), names of patients who happened to be present, or meaningless words, were produced with the tic. In the fifth case (A. Sh—) the connection was especially striking. The patient developed " St. Vitus' Dance " after being knocked over by a dog. The abnormal movements varied in severity and were described as occurring in attacks, but she was probably never entirely free from tic-like movements thereafter. They involved mainly the face and shoulder, and were accompanied by grunts and sniffs. At 24 she became illegitimately pregnant, and during a septic illness following delivery her movements were, for the first time, accompanied by coprolalic utterances bearing an unmistakable sexual connotation.

Comparing our cases, we see how the identical motor impulse is filled with contents depending on the actual situation. Osler (12), as long ago as 1894, described a similar sequence of events in the case of a child who first showed spasmodic movements, later meaningless utterances, and eventually compulsive utterances of an obscene kind, which later disappeared. Straus has rightly pointed to the analogy with the swearing of normal people, where the motor verbal eruption is the essential thing. The words used are unimportant ; they may be mutilated and deprived of their proper sense, but the motor design, the motor Gestalt as it were, the rhythm and the pitch, remain. In contrast to Straus's assumption, we did not find that the struggle against the forced movements affords the emotional impetus for the coprolalia. Our cases tolerated the hyperkinesias surprisingly well, as do many patients with chorea. They experienced the utterances, however, as something involuntary, unfamiliar, and as if forced upon them.

Meige and Feindel (13) have said that " the frequency with which obsessions, or at least a proclivity for them, and tics are associated cannot be a simple coincidence ". These authors are, therefore, inclined to derive one symptom

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from the other, to conceive of either the tic as the motor reaction to an obsessional idea, or the obsession as an answer to the tic. Modern writers, Wilder and Silbermann (14) for example, observing the coincidence of the above symptoms in chorea and encephalitis, have given more attention to the complicated interaction of psychic and physical factors. In this connection we would refer again to Straus, who saw in the motor impulse the essential source of the compulsive utterances.

In the cases which Meige and Feindel have in mind, permanent obsessive features are reported in tiqueurs rather than attacks of obsessive-compulsive phenomena. It is only since observations have been made on encephalitis that attention has been focussed on the paroxysmal character of such obsessive-compulsive phenomena.

Oculogyric crises are ranged alongside the tics by some authors. In doing so, one particular difference stands out among the many similarities; the cases with oculogyric crises tend to have obsessional thoughts, the respiratory tics tend to have compulsive utterances. That this connection is regularly observed suggests an internal relation. Kleist (15) and others have observed disturbances of eye movements associated with disturbances of attention. Calling the oculogyric crises "spasms of attention" gives a hint as to their significance.

The only instance given by Meige and Feindel of a paroxysmal obsession was in a case compelled to count three while performing her tic, which consisted in having to look *up* to the curtains in a corner of a window. Here, in fact, we see just such an instance of a compulsive eye movement, associated with a recurrent obsessional thought. This gives rise to a working hypothesis, requiring much further observation and corroboration, to the effect that the groups of muscles affected by the tic may determine the nature of the accompanying psychic phenomena.

The cases of chorea which tend to persist or go on to tic seem to show some differences from more typical Sydenham's chorea at quite an early stage. Among such differences may be a preponderance of involuntary choreiform movements in the region of the face, especially the mouth, and for the muscles of respiration, phonation and articulation to be involved—a type of chorea first singled out by Oppenheim—thus laying the track, or facilitating the later symptom of vocalized tics. Further, some of the cases of chorea reported in this connection show signs of deterioration suggestive of widespread organic involvement. This may constitute a genuine *maladie des tics*, around which are grouped the divergent symptomatic forms.

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Case Records.

Case 1.—G. R—, a man, æt. 22, had chorea at the age of 7. His childhood was otherwise uneventful. He left school from Standard VII, and obtained work as a packer. Later he enlisted in the Royal Marines and saw active service in China.

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He was invalided out with a second attack of chorea which occurred after being in a bombardment. A third attack occurred eighteen months before admission, during a period of unemployment. He is said to have been discharged from his last place of work because of his abnormality. On admission he showed jerking movements of his face and head, with a grunting respiratory tic. These movements were spasmodic, and were more in evidence when he was embarrassed. He improved during his stay in the ward.

CASE 2.—A. V—, a boy, æt. 15, of average intelligence, showed spasmodic movements of face, limbs and respiratory muscles, resulting in meaningless noises, but no verbalized utterances. The movements vary, gradually decrease during an interview, may cease entirely before strangers, but tend to increase on first changing into new surroundings. Face, lips and tongue move irregularly, with a brisk, tic-like quality, and expiratory grunts occur as the patient attempts to check these movements. His mood is usually cheerful and irresponsible. Some degree of motor instability is suggested by the marked spread of associated movements, and Gordon's phenomenon is present. The movements can for a time be controlled by the patient, but screening showed irregular contractions of both diaphragm and thoracic muscles. Some grunting occurred without corresponding movements of diaphragm or chest. No compulsive features were elicited.

The duration of these symptoms was six years. The family history is negative, and his previous health has been good except for several "attacks" of chorea, the first when 8 years old. When 13 he spent six months at Queen Mary's Hospital, Carshalton, sent in as a case of chorea, but regarded there as mainly unstable and debilitated. The movements were present at first, but ceased during his stay. At that time no vocalized tics were noticed, and there have been no rheumatic sequelæ.

CASE 3.—A. S—, an intelligent boy, æt. 10, had for a month shown movements resembling chorea, mainly affecting face and shoulders, following a sore throat. Recently he is said to have muttered and repeated remarks under his breath. The family history is positive, one elder sister having had two attacks of chorea, both observed by one of us, while another sister had a period of timidity accompanied by involuntary movements. In this boy rheumatic ætiology is suggested by a history of pain in the shins, and the onset after tonsillitis. When seen, in addition to movements of the face, tongue and shoulders resembling chorea he showed certain regular twitchings of the face and shoulder resembling a tic. No "muttering" was observed, but the history was probably reliable. Recovery appeared to coincide with the exhibition of salicylates and a short period away from school; there has been no recurrence on return there.

CASE 4.—M. S—, a girl, æt. 23, is now a voluntary patient in a county mental hospital. Her present symptoms are slight; sometimes she is restless or fidgety, calls out names or words without knowing why, and she has impulses to disfigure bedding or clothes, especially with soot or blacklead. She is never incontinent, and there is no serious dementia, but she remains unambitious and does not resent her present surroundings. The previous history includes attacks of chorea at 8 and 11 years old. She was always a nervous, shy child, but was regarded as of normal mentality. The family history is negative and there is no evidence of rheumatism.

When she was 18 she attended the Maudsley Hospital Out-Patient Department. Choreiform movements were then present, with spasmodic grunting noises, also the compulsive features of having to soil or destroy clothes. Her intelligence at that time was normal, on a Burt revision of the Binet-Simon test.

This is now seen as a slowly progressive case, the gradual onset of present symptoms dating from when she left school.

CASE 5.—A. Sh—, a single woman, æt. 32, with a tic involving face, shoulder and limbs. Jerky spasmodic contractions are accompanied by occasional grunts or sniffs, and sudden voluntary movements, such as flinging her hand behind her

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back. The left arm and leg are more affected than the right. The movements never entirely cease, but the frequency and severity are variable.

The onset of abnormal movements was at 13 years, when, after being knocked over by a dog while running, she lost consciousness. This was followed by " St. Vitus' Dance ", varying in severity, but apparently continuing until she was 16 years. She was never entirely free from movements thereafter. When she was 27 she became illegitimately pregnant, went to full term, and had septicæmia after delivery. Movements had practically ceased during the second half of her pregnancy, but recurred severely during the septic illness, accompanied for the first time by compulsive utterances of an obscene kind. She improved, and there was a subjective sense of lessening in severity of tic, though little change was noted by observers. Now her mood is euphoric, notably in her delight in achieving a pregnancy. She is inclined to posture and attitudinize, apparently without self-consciousness relating to her obvious and severe tic.

CASE 6.—D. C—, a boy, æt. 10, has had choreiform movements of varying severity for five years. For the last five months only he has been noticed to mutter irrelevant obscenities as if compelled to do so. Movements are mainly of the face and lips, with irregular respiration, grunts and " clucking " noises with lips and tongue; there is some tic-like grimacing, regular and repetitive. That the patient is unaware of some of the movements is suggested by the choreic position of extended hands, fine movements of the fingers, and an exaggerated spread of associated movements, while X-ray screening showed, as in Case 2, irregular contractions of the diaphragm. Many neurotic features are present, such as anxiety, incontinence, fear of the dark, and nail-biting.

The onset was at 5 years, two months after a shock, when he was attacked and mauled by a dog. He has missed much school, and is, therefore, backward. He has often complained of pains in the joints, and is looked on as being delicate. There are no signs of active rheumatism at the present time, and the family history is negative.

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