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Gilles de la Tourette Syndrome and Mania in an Adolescent

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Summary: The case is presented of a twelve-year-old boy suffering simultaneously from Tourette Syndrome (TS) and a manic episode. The problems of this unique clinical association are discussed from the point of view of differential diagnosis, treatment, and possible biochemical basis. The case also illustrates the usefulness of the expanded concept of TS as a neuropsychiatric disorder with dysregulation of perceptual, motor, and cognitive functioning.

Tourette syndrome (TS) is a severe and chronic neuropsychiatric disorder, which usually begins in childhood, and is of uncertain aetiology.

Since the condition was initially described in the nineteenth century by Itard and George Gilles de la Tourette, the accepted essential clinical features of multiple motor and vocal tics remain the mainstay of the diagnosis (Silver, 1980). Nevertheless, clinical observations over the years have shown that many of these patients have a large number of associated psychological and behavioural symptoms and signs, such as attentional difficulties, hyperactivity, obsessive thoughts and compulsive rituals, learning difficulties, impulsive an anti-social behaviour, inappropriate sexual activity, and sleep disturbances (Cohen *et al,* 1980; Nee *et al,* 1980; Shapiro *et al,* 1978; and Silver, 1980). Some of these features (attentional difficulties, hyperactivity, obsessive compulsive symptoms) may appear before the tics develop, so that they cannot be regarded as secondary phenomena (Cohen *et al,* 1980).

Thus TS may be conceptualised as a neuropsychiatric disorder with dys-regulation of cognitive, perceptual and motor functioning. The tics merely represent one aspect of it, possibly the least disabling (Bliss, 1980; Cohen *et al,* 1980). There have been attempts to explain the disorder in terms of a psychotic illness, but these postulates have not been substantiated by clinical evidence. Epidemiological studies have not shown an increased prevalence of psychosis in TS patients, compared with controls (Shapiro *et al,* 1978).

We present here a case of TS and mania coexisting in an adolescent boy. Both these conditions are difficult diagnostic problems for the clinician when they occur in adolescence, especially when they appear together.

Case report

L is a twelve-year-old boy in early puberty, of Jewish extraction. He is the third of four children in a family of low socio-economic status. There is no family history of psychiatric or neurological disorders, and the nuclear family itself is a warm and caring one.

Early psychomotor development was normal until the age of three, when he was diagnosed as suffering from 'minimal brain dysfunction' with hyperactivity, attentional difficulties, low tolerance to frustration, and emotional lability. He was referred for special education, but after one year was returned to the regular school programme, where despite his average intelligence, he showed poor academic performance.

At the age of eight, he began to suffer from obsessional thoughts as expressed by 'nonsense words', which stuck in his mind and which he could not get rid of, however much he tried. He began to force his parents, especially the mother, to perform compulsive rituals such as giving standard answers to standard questions. When they did not comply, he would lose control and go into a rage. At the age of ten, he developed—in addition to the above symptoms—facial and head tics, spreading to the upper limbs, with concomitant deterioration of his behaviour expressed by temper tantrums, uncontrollable spitting, and use of obscene language.

At this stage, he was hospitalised in a children's psychiatric unit, where he was diagnosed as having "conduct disorder secondary to nonspecific brain dysfunction". During the following two years, his behaviour problems became progressively worse, despite attempts to treat him with psychotherapy, anticonvulsive drugs, stimulant medication, tricyclic antidepressants, and various antipsychotic agents.

His rage attacks became so uncontrollable that he had to be transferred to our locked unit, after which all medication was withdrawn and the boy was observed for a period of six weeks. During this time, he developed vocal tics, which expressed themselves as animal-like sounds, and also typical features of manic episode including euphoric mood, his constant laughing, facetiousness and singing of football songs turned to anger, with physical violence, at the slightest provocation. He was hyperactive, unable to sit still, continually provoked other children and had marked difficulty of concentration. He showed pressure of speech and flight of ideas, with delusions of grandeur ("I am the world's greatest soccer player, I am the world's champion in chess, I am the handsomest boy in the world..."). Physical, neurological, electroencephalographic and computerised tomographic examinations were all normal. In view of the patient's history and mental state, we arrived at the two separate coexisting diagnoses, *viz*- of Tourette Syndrome and affective disorder (manic type).

Due to the complexity of the clinical problem and in order to avoid polypharmacy if at all possible, we decided

to initiate treatment only for the Tourette Syndrome. Therefore the patient was given Doparid (Tiapride), a member of the substitute benzamide series that interacts preferentially with dopamine DA_2 receptors (Buruma *et al*, 1982; Dalery *et al*, 1980), up to 300 mg daily for 18 days, without any significant beneficial effect. We then instituted a trial of clonidine, an alpha adrenergic agonist which reduces noradrenergic activity (Cohen *et al*, 1979) in doses of up to 0.25 mg a day for 24 days. This led to reduction of the facial tics, but the vocal tics, manic symptoms, temper tantrums, and the obsessional thoughts did not improve at all. We subsequently switched to haloperidol seven milligrammes daily. The patient's tics, which had reappeared in the washout period after stopping the clonidine, disappeared completely; the manic symptoms improved greatly, and the behaviour problems improved, though they did not remit completely. The patient continued to suffer from obsessional thoughts, but in a milder form than formerly.

Discussion

This case illustrates the complexity of the clinical problems posed by Gilles de la Tourette syndrome, but the enlarged concept of the disorder suggested by Cohen *et al* (1980) and others enables the clinician to make the most succinct definition of a complex clinical picture, and thus to devise more rational approaches to treatment.

In contrast to Tourette Syndrome, which is relatively rare (incidence 0.1 to 0.5 per 1000, Silver, 1980), affective disorders are being increasingly diagnosed in childhood and adolescence. It is now estimated that 20 to 35% of bipolar illness begins during or before adolescence (Loranger & Levine, 1978). The criteria for mania in childhood, such as those of Weinberg & Brumback (1976) are: (i) euphoria (ii) irritability (iii) hyperactivity (iv) pressure of speech (v) flight of ideas (vi) grandiosity (vii) sleep disturbances and (viii) distractability. These show considerable overlap with the symptoms of Tourette Syndrome as described earlier, making the differential diagnosis of the case presented here all the more difficult.

It is possible to speculate on an overlapping of the biochemical bases of these conditions. The catecholamine hypothesis of affective disorders states that mania is a result of excessive noradrenergic activity in the central nervous system. Clonidine is an alpha adrenergic agonist which reduces CNS noradrenergic activity and has also been shown to be effective in the treatment or TS (Cohen *et al*, 1979, 1980); thus, we may postulate that TS, like mania, reflects excessive adrenergic activity in the brain. This is supported by the similarity of the clinical picture of both disorders, and by their coexistence in one patient, as described above. A factor militating against such a speculation, however, is our patient's relative lack of response to clonidine.

It should be noted, though, that he had received prolonged stimulant medication therapy in the past, which Cohen (1980) feels may interfere with the success of later clonidine therapy in TS.

Indeed, the long-term effects of the many and varied drug combinations which this boy had received may well have contributed to the clinical picture. Tourette syndrome itself may develop as a complication of antipsychotic therapy (Klawans *et al*, 1978); though this seems unlikely to have been the case with our patient, since most of his clinical features developed from an early age.

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