British Journal of Psychiatry (1986), **149**, 644–647

Brief Reports

A Clinical Study of Gilles de la Tourette's Syndrome in Korea

Clinical data on 30 Korean patients of the authors with Gilles de la Tourettes's syndrome are described, as well as data on seven other Korean cases from the literature. The overall characteristics and EEG findings were similar to those of Western patients, but there were fewer obsessive-compulsive behaviour problems, and family histories were less often positive.

Gilles de la Tourette's syndrome is an unusual condition, characterised by the onset in childhood or adolescence of recurrent, involuntary, repetitive, and rapid movements which include multiple vocal tics. Coprolalia, copropraxia, echo-phenomena, and obsessive-compulsive traits may also be present, but are not diagnostic prerequisites. Aetiologically, it has been considered at least partly a psychogenic disorder, but neurophysiological studies suggest that its basis is neurological (Shapiro & Shapiro, 1982), while Lees *et al* (1984) have reported evidence for heterogeneity.

Although many cases have been reported in western countries over the past 100 years, there have been fewer than fifty from Asia in the English-language literature. Only about ten reports of Asian cases could be found in the Australian MEDLARS and in Index Medicus. Although one was reported in India in 1962, the first Asian case is generally thought to be that of a Chinese boy in Hong Kong (Singer, 1963). The scarcity of such reports does not necessarily reflect the rarity of the syndrome, but rather the lack of awareness of it, and failure to publish cases in English-language journals (Lieh-Mak *et al*, 1978).

In Korea, six cases have been published since Lee & Sul (1968) reported the first case (that of a seven year-old girl); all of these were in Korean medical journals only. Suh & Park (1969) reported a 14 year-old boy with the syndrome who had suffered from sibling rivalry and a domineering and punishing father. Kim (1971) described a ten year-old boy, in whom the aetiological factors were said to be hostile identification and ambivalence to a father figure and an unconscious desire for self-punishment. Koh *et al* (1971) reported a 20 year-old male, whose typical symptoms improved with phenothiazine. Rhee & Kim (1973) described a 27 year-old male, in whom the aetiological problems were postulated as hostility and poor impulse control towards authority figures; he had also a typical obsessive-compulsive disorder, and haloperidol was used for the first time in Korea in this case. Kim & Lee (1973) reported a 20 year-old female, and Kang & Yoon (1974) a 14 year-old boy who has had phobias and compulsive behaviours; psychodynamic factors were described as a tyrannical father figure and feelings of rejection.

In this report, we summarise the clinical data on 30 Koreans with the syndrome whom the authors have seen personally since 1976, along with additional data from the seven cases described above.

Method

Thirty patients were diagnosed as having Gilles de la Tourette's syndrome during the period 1976–1984 at the Department of Psychiatry, Yonsei University Medical Center, Seoul. All were evaluated psychiatrically by one of the authors; history of symptoms and family and developmental history were also obtained from semi-structured interviews with the patients and their families.

For final inclusion, all clinical data were reviewed again by both authors for the following diagnostic criteria: onset of symptoms between the ages of 2 and 15; presence of recurrent, involuntary, repetitive, rapid, purposeless motor movements, affecting multiple muscle groups; and multiple vocal tics. Ability so suppress movements voluntarily for minutes or hours, variations in the intensity of symptoms over weeks and months, and duration of more than one year, as required by DSM–III (American Psychiatric Association, 1980), were not considered necessary for the diagnosis; neither was coprolalia.

Twenty-nine patients met these criteria. In a further female patient, the syndrome began at the age of 18. Her symptoms, such as barking and multiple tics, were typical of the syndrome, had been present for about four years, and were controlled by haloperidol. She was therefore diagnosed as having Gilles de la Tourette's syndrome, and included in the study.

Eight of the cases were referred by neurologists, neurosurgeons, paediatricians and physicians, and nine by other psychiatrists in the same department.

Results

All 30 patients were native Korean citizens. The syndrome appears to be rare in Korea, considering that the total number of out-patients of this Medical Centre was about four million during the period. Twenty-four patients were males and six females. The average age was 11.8 years, and ranged from 6 to 32; 24 patients were in the range 8–16 years. One of the four adults was married.

No significant factor was reported in the birth history of any patient, but 15 had an abnormal developmental history, including hyperactivity and distractability in nine cases, learning difficulties in four, enuresis in two, encopresis in one, and stuttering in two. Twelve patients were reported by parents to have irritable and impulsive personalities, eight to be competitive and egocentric, five to be obsessive and meticulous, and five to be shy and polite.

As far as the medical history was concerned, two patients had suffered from nephritis, two from dog bites, two from pulmonary tuberculosis, one from a car accident, and one from febrile convulsions. One patient had had a tonsilectomy. These previous problems did not seem to play a role in the initiation of the symptoms.

Ten patients were the youngest of their families, six were in between more than three siblings, four were oldest sons, and eight were only sons or daughters. Simple tics were reported to have been present in the siblings of only two patients. The presence of functional psychosis in first-degree family members was denied in all cases except one, where the grandmother died of an unspecified psychosis.

The syndrome began before the age of 15 in 29 cases and at age 18 in one female, who can be considered as an example of an adult-onset type of the syndrome. The mean age of onset was 8.7 years for males and 7.8 for females, while the commonest age of onset was between two and four years (eight patients). The mean duration of the illness was 3.8 years (range 0.1–22 years).

Various precipitating and situational factors might reflect psychodynamic influence on the development of symptoms, e.g. tonsillectomy in two cases, and beating by the father, elder brother, or schoolteacher in four cases. Eleven other cases were thought to show psychogenic factors, including emotional frustration, conflicts in child-parent relationships, and loss of self-esteem; two patients scarely showed the symptoms at school, but frequently did at home.

Most patients reported that the symptoms, especially vocalisations, were usually aggravated by anxiety, tension, and other emotional stress. Fifteen reported anxiety, emotional uneasiness, and chest tightness when they tried to voluntarily control or inhibit symptoms – especially with coprolalia in four cases. Common cold with fever seemed to be related to recurrence in six cases.

After the initial symptoms, other parts of body became involved, and new tics developed which replaced or added to pre-existing ones; the general progression was cephalocaudal. These symptoms are shown in Table I. Obsessive-compulsive personality traits were noted in five patients, but such behaviour, e.g. touching and changing clothes, was reported in only three of these.

Only nine cases showed the classical syndrome with multiple motor tics, barking, and coprolalia; another eight had combined multiple motor tics and barking. Nine cases had multiple motor tics and other vocalisations, while four had only severe multiple motor tics. According to the criteria of severity of Shapiro *et al* (1973), 14 patients (47%) were rated as having marked and severe illness, i.e. progressive and chronic, without remission, and with serious social, academic, and occupational impairments.

No patients were evaluated as having schizophrenia or other psychoses. Ten children were reported to have

TABLE I

Initial and presenting symptoms in 30 Korean cases with Gilles de la Tourette's syndrome

Symptoms	*Number of patients*	
	Initial	*Presenting*
Eye twitchings	15	16
Tongue movements		3
Grimaces	4	9
Barking		14
Other vocalisations	4	9
Head and neck jerks	6	20
Arm jerks		12
Leg jerks		10
Trunk jerks	1	5
Coprolalia		9
Spitting		5
Echolalia		4
Compulsive behaviours		3
Stuttering		2
Bizarre mannerisms		2

marked adjustment reactions to peer relationships at school since the onset of the syndrome, e.g. one eight year-old boy refused to attend school because of teasing by peers. Two patients had suffered from nightmares. Four patients, who were older, were diagnosed as having reactive depression; one of them, a 17 year-old male, showed serious depression with suicidal ideas. The parents of six children reported that the personality of their children had changed since the onset of the syndrome to become hyperactive, violent, and impulsive.

Thirteen of 25 patients who had EEG examinations showed non-specific abnormal findings: random slow waves in three cases, random slow and sharp wave mixtures in five, mild focal abnormality in one, and a burst of high amplitude sharp waves in both hemispheres in four. Computerised tomography was carried out in six cases, and the findings were normal in all these.

Among previous treatments were anticonvulsant medication in five cases diagnosed as having a myoclonic epilepsy before referral, benzodiazepines in three, herbal medicine in five, and acupuncture in two. One case tried treatment by traditional Korean exorcism. All these treatments eventually proved to be ineffective.

After the diagnosis of Gilles de la Tourette's syndrome, all patients were treated with haloperidol. The initial dose was usually 0.5–3.0 mg/d, according to age and body weight, gradually increased by 0.5 or 1.5 mg until an effective dose was determined. This was different in each patient, and ranged from 1.0 to 12.5 mg, with a median of 2.5–3.0 mg/d; it did not seem to be significantly correlated with the body weight, age, severity of the symptoms, duration of illness, or age of onset. The period of treatment ranged from three weeks to three years.

With haloperidol, symptoms remitted completely in 21 patients and were partially alleviated in five others, after about one month; this was evaluated by clinical judgement of overall percentage decrease of frequencies of symptoms,

compared with the pre-treatment levels. The average improvement rate was 87%. Combined treatment with diazepam (3–10 mg/d) was helpful in five cases in which emotional tension was an obstacle to treatment. The side-effects of haloperidol were troublesome to many patients: diurnal drowsiness was most common, while acute dyskinesia was the major cause of discontinuing haloperidol.

Twenty-five patients discontinued medication against medical advice, after 3–12 weeks of treatment. In most cases, either the patients or their parents believed that treatment had been sufficient, but 17 patients visited the hospital again after a mean period of 2.6 months because their symptoms recurred; in four of these, the dosage needed to be increased to obtain the same effect as given by previous treatment.

Discussion

In this study, the clinical features of Gilles de la Tourette's syndrome such as age of onset, duration of illness, male predominance, non-significance of birth rank, and relatively high incidence of abnormal developmental problems revealed no differences from those in previous studies on European, American, or Asian patients (Singer, 1963); Morphew & Sim, 1969; Shapiro *et al*, 1973; Woodrow, 1974; Fernando, 1976; Lieh-Mak, 1979; Nomura & Segawa, 1982; Chen & Lu, 1983; Lees *et al*, 1984). The frequency of first symptoms, cephalocaudal progression of symptoms, waxing and waning clinical course, frequency of involved muscle groups, and incidence of EEG abnormality were also similar. The seven cases reported by other Korean authors had clinical features almost the same as in this report: the typical syndrome with coprolalia was found in two cases, and multiple motor tics (including vocal) in four. These findings suggest that the clinical history and general symptom pattern of Gilles de la Tourette's syndrome, although rare, are almost uniform throughout the world.

However, there were minor differences: in this study, neurotic traits were not reported as often as in Western series. In particular, obsessive-compulsive personality traits or behaviour problems were less frequent than in the cases reported by Morphew & Sim (1969), Woodrow (1974), and Lees *et al* (1984). Other Korean authors have reported one case with a history of compulsive behaviour and two cases with obsessive-compulsive personality traits (Rhee & Kim, 1973; Kim & Lee, 1973). However, Shapiro *et al* (1973) concluded from 34 European and Jewish patients and from the literature that obsessive-compulsive symptoms and traits are infrequent. The lesser incidence of tics or other psychiatric problems in the family histories of this series and of other Korean cases contrasts with the high incidence in Western studies, which had provided a genetic model for the syndrome (Kidd *et al*, 1980). There are two possible reasons for this: intentional denial by Korean parents, because of the prejudice against psychiatric illness; and failure to use a standardised rating scale to detect them.

The cause of the syndrome is still obscure. Although there is a trend to believe that it is a neurological disorder of the central nervous system (Shapiro & Shapiro, 1982), supported by the presence of non-specific abnormal EEG findings in half our cases, Korean authors have attempted a psychodynamic formulation. Hostile father figures and inhibited aggression (Kim, 1971; Rhee & Kim, 1973; Kang & Yoon, 1974) have been postulated as core problems; similar findings occurred in some cases of this series. Tics, particularly coprolalia and spitting, might indicate a disturbance in normal balance between a need for tension relief and the capacity to control such actions, suggesting that psychogenic factors may be aetiologically relevant (Ascher, 1948; Dunlap, 1960).

Haloperidol was as effective a drug for Korean patients as it is for Caucasians and other Asians, but the dosage must be individualised; the average improvement of 87% was similar to the rates reported in western countries (Bruun *et al*, 1976; Woodrow, 1974). Three of the other seven Korean cases were also treated with haloperidol, while another two were treated with perphenazine and chlorpromazine. All these drugs were effective, and their dosage was decreased gradually. However, our study suggests that Korean patients need less haloperidol than Caucasians, who had 6–180 mg/d, frequently in addition to anti-parkinsonian drugs (Shapiro *et al*, 1973). However, the same authors (Shapiro & Shapiro, 1981) have reported responses to remarkably low doses, e.g. 5 mg daily. Asians may have a genetically different constitutional responsiveness to drugs: Nomura & Segawa (1982) reported that haloperidol (0.75–3 mg/d) was effective for about 70% of 100 Japanese, Korean, and Philippine children with the syndrome, while Chen & Lu (1983) reported that haloperidol (0.5–3 mg/d) showed excellent to fair improvement in eight out of ten patients.

Our findings confirm that the Gilles de la Tourette's syndrome is seen in Korea, and has many features in common with those reported from Western countries. The aetiology probably represents multiple factors, which need to be understood in terms of emotional, behavioural, developmental, hereditary, and organic concepts.. Evaluation and treatment should encompass a holistic approach, and include chemotherapy, academic help, counselling, and other psychosocial considerations.

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(*Accepted 4 July 1985*)

British Journal of Psychiatry (1986), **149**, 647–650

The Life Satisfaction Index – Well-being: Its Internal Reliability and Factorial Composition

The internal consistency and factorial composition of the eight-item Life Satisfaction Index – Well-being, adapted for use with elderly British samples, were examined. Cross-validation was carried out with a randomly drawn community sample of 155 rural people aged 65-89 years. The scale was found to have acceptable internal reliability, but may require modification.