GILLES DE LA TOURETTE'S SYNDROME: A STUDY OF THE TREATMENT OF SIX CASES BY MASS NEGATIVE PRACTICE AND WITH HALOPERIDOL

By J. I. Teoh

SYNOPSIS

Six cases of Gilles de la Tourette's Syndrome were described. The technique of mass negative practice causing reactive inhibition was used to treat two cases. Contrary to previous studies, mass negative practice reduced the quality of tics but did not affect the frequency of tics in the syndrome. Subsequently all the cases were treated with Haloperidol with a dramatic remission of symptoms. The video-tape method of measuring percentage improvement of symptoms was described.

INTRODUCTION

This unusual form of multiple, generalized tics accompanied by explosive utterances was originally described in 1825 by Itard and later classified as a syndrome by Georges Gilles de la Tourette (1885). The symptoms considered diagnostic of the syndrome are:—

(a) Childhood onset (usually below 16 years)
(b) Multiple motor tics, and
(c) Unprovoked loud utterances, which may progress to compulsive shouting of obscene language (corprolalia).

The illness usually commenced with multiple motor tics sometimes accompanied by utterances (vocal tics). According to Fernando (1967) the more common tics were motor tics affecting the head, face and neck, while the less common were limb and trunk movements. Other tics, like grimacing, teeth grinding and tongue protusion were sometimes reported, but more complex movements like the hitting of the face or genitalia were rarer. Fenichel (1945) observed the sexualization of speech and interpreted it as a form of compulsive neurosis.

The prognosis has been described by authors (Chapel *et al*, 1964) as "sinister", uniformly poor and "unresponsive to therapy", with a relentless clinical course marked by progressive and marked personality changes which may last through the life of the individual. Faux (1966) described the tragic life-long institutionalization of such cases where treatment previously had been mainly socio-psychiatric methods. However, several authors (Heuscher, 1953; Bockner, 1959) reported spontaneous remissions in late teens and twenties lasting over several years.

The illness has a marked social impact and patients often sought help for social, personality and behavioural difficulties. Their behaviour often deteriorated, when treatment failed and frequently psychopathic behaviour resulted.

AETIOLOGICAL FACTORS

(a) Psychological Theories

The aetiology of the syndrome is largely unknown and the diagnostic label "psychogenic" is often used. However, there is very little agreement about psychological factors common to patients, except for frequent reports of compulsivity and inhibited aggression. An underlying psychodynamic cause of a suppressed hostility to parents or significant object-relations was suggested by several authors (Ascher, 1948, Eisenberg *et al*, 1959, Dunlop, 1960 and MacDonald, 1963). Likewise, Fernando (1967) reported that 57% of all cases surveyed had marked obsessional tendencies, and the majority had some precipitating pyschological stress (Morphew *et al*, 1969).

Otto Fenichel (1945) interpreted the corprolalia as having a strong anal-sadistic component and that the utterances of obscenities were an aggressive act directed at the listener. He postulated that there was a "magical" temptation to utilize obscene and profane words to attack the listener violently or sexually.

(b) Organic Theories

Although there is no evidence of a consistent association between the onset of tics and any physical illness, and no organic aetiology can be definitely attributed, the clinical data suggests that

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the condition is a bizarre and rare organic neuro-physiologic impairment of the central nervous system (Corbin, 1968, Wagner, 1970 and Shapiro *et al*, 1971). All that is known is that the basal ganglia may be the probable site of disturbance. The only positive autopsy case (Claus and Balthason, 1954) demonstrated an immature cell structure of the corpus striatum. Generally, positive neurological findings are rare in all reported cases, although abnormal EEG findings were reported in twenty-five percent of all cases analysed.

(c) Neuro-Physiological Theories

Several lines of evidence (Synder *et al*, 1970) suggest that symptoms of the disease may be related to brain dopamine in the corpus striatum. The dramatic therapeutic responses (Shapiro, 1968) obtained with the butyrophenone Haloperidol give some clues to the neuro-physiology of the syndrome. Phenothiazines and butyrophenones markedly accelerate the turnover of dopamine in the corpus striatum. This mechanism of accelerated turnover is thought to be due to the blockade of dopamine receptors in the corpus striatum, causing an enhancement of a postulated feedback to the pre-synaptic dopamine neurones, which respond by increasing dopamine synthesis, (Thus, both the symptoms of idiopathic and drug-induced parkinsonism can be attributed to a relative deficiency of dopamine at receptor sites in the corpus striatum).

The fact that Haloperidol is unique in its great potency for blocking dopamine receptors and that it is effective in Gilles de la Tourette's Syndrome may account for a patho-physiology of the condition. It is proposed that in the corpus striatum of such patients, there is a hyperactivity of dopaminergic systems. Whether this is produced by enhanced release of dopamine, impaired inactivation of dopamine, or hypersensitivity of receptors is a matter for speculation. The proposed hypothesis would account for the resemblance of symptoms of Gilles de la Tourette's Syndrome to the side-effects of l-dopa therapy. This would also explain the specific therapeutic efficacy of Haloperidol.

CASE STUDIES OF GILLES DE LA TOURETTE'S SYNDROME

Six cases of Gilles de la Tourette's Syndrome (see Table I) are discussed and the following histories are described.

Case 1

A 35-year-old Malay male presented a history of facial tics since six years old. By 11 years old, he had developed facial grimacing and pouting of the mouth. Head nodding started at 21 years old followed by obscene utterances. He married at 30 years old and his symptoms became more severe after the birth of his second child. They involved tics of both upper and lower limbs, obscene gesticulations, grunting noises and compulsive shouting of the "four-letter word". He started stamping his feet, slapping his face and hitting his genitals uncontrollably. His wife became so upset that she divorced him and promised to return only if he sought a cure for this strange malady.

During the time of consultation, he had been unemployed for several years and had by then gone all over the country including Sumatra in search of a cure to his illness.

TABLE I

CHARACTERISTICS OF PATIENTS WITH GILLES DE LA TOURETTE'S SYNDROME AND MAXIMUM DOSES OF HALOPERIDOL AND BENZHEXOL/DAY DURING THE TEST PERIOD

	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6
Ethnic Group	Malay	Chinese	Chinese	Chinese	Chinese	Chinese
Sex	Male	Male	Male	Male	Female	Male
Age in years	35	19	25	22	16	23
Max. dose Haloperidol/day	19·5 mg.	37·5 mg.	33·0 mg.	6·0 mg.	9·0 mg.	12·0 mg.
Max. dose Benzhexol/day	32 mg.	36 mg.	24 mg.	12 mg.	12 mg.	16 mg.
Duration of Test period	90 days	27 days	33 days	30 days	28 days	28 days

Case 2

A 19-year-old Chinese male, a fishmonger, developed the illness at eight years old when he felt an urge to clear his throat frequently. This progressively worsened as he started producing grunting noises. By 14 years old, he felt a compulsive urge to frequently hit his abdomen with his hands. Two years later, he started grimacing and shouting out obscene names of his friends repetitively. He left school at this juncture because of his anti-social symptoms. By the time he reached 17 years old, he was compulsively shouting obscenities, indecently gesticulating with his hands and repeatedly stamping his feet. He eventually gravitated his corprolalia to women essentially and this behaviour was reinforced when he was assaulted by their husbands.

He gave a history of having frequently observed his parents performing coitus, when he was seven years old. He developed hostile feelings towards his father and would attempt to sleep in-between his parents to prevent coitus and deliberately made throat sounds to distract his parents. His symptoms appeared to have been precipitated by witnessing the primal scene.

By the time he was admitted to hospital, he was depressed, ashamed of his symptoms and harboured strong sado-masochistic tendencies.

Case 3

A 25-year-old Chinese male, a motor-car mechanic, developed insidious symptoms of coughing at 16 years old. His concentration deteriorated and he developed insomnia. Very soon he started cursing in an explosive manner and compulsively cursed obscenities directed mainly at his mother. He felt more relaxed after corprolalia. Later he began gesticulating obscenely with his hands. The symptoms became progressively worse, with echolalia, corprolalia and indecent gesticulations several hundred times per hour. He would hit his testes until they hurt and stamp his feet repeatedly. Eventually he learned to distort his obscene corprolalia by swallowing the "four-letter word" and by sucking his thumb. This resulted in pressure headaches which he suffered for many years.

He blamed excessive masturbation for his illness and was unable to stop the habit which he indulged in three to four times per day. The symptoms had a devastating social effect on his life. He isolated himself socially, avoided female company and likewise female clients avoided him. He felt very ashamed of his symptoms, which he was unable to control and became very depressed and desperate during the time of admission to hospital.

Case 4

A 22-year-old Chinese male, a college student, gave a history that at the age of seven years old, he witnessed his parents performing coitus. This traumatic experience made him hate his parents and he became depressed each time he spoke to them and painstakingly avoided staying at home. He became so obsessed by the sexual act that he ruminated over the incident even until the time of consultation. Since then, his relationship with his parents has been cold and distant. This incident coincided with a female neighbour, whom he was infatuated with, and who had left the neighbourhood.

Soon after that incident, he started coughing and making queer noises in his throat. This was followed by the wrinkling of his nose, the repeated winking of his eyes and active nodding of his head. At times, while walking, he would stamp his feet. These symptoms developed gradually and fluctuated in intensity. Occasionally he would compulsively blow his nose and make semi-purposeful movements.

His symptoms interfered with his studies, caused him untold embarrassment and he socially isolated himself. His concentration deteriorated and he became nervous and depressed by this uncontrollable disorder.

Case 5

A 16-year-old student, a Chinese female, was brought by her father who noted that she had insidiously developed fidgetiness at 13 years old. Initially she developed a blocked nose and started a sneezing habit, with a twitching of the nose. This subsequently spread to involve mouth twitching and grinding of her teeth. One-and-a-half years later she developed jerky limb movements and was unable to control hitting herself.

Her symptoms increased and she became withdrawn. Her self-hitting became more severe and spread to hitting her mother and grandmother. Frequently, she would make grunting noises and swear curse words under her breath and then feel most embarrassed and repentant. During the time of admission she had obsessive-compulsive rituals, corprolalia, echolalia, stamping of feet and had learnt compensatory movements to dampen her gesticulations.

Her father described her as a reticent girl who fared poorly at school. He had taken her to approximately 100 doctors, native healers and mediums in a search for a cure to this disturbing malady.

Case 6

A 23-year-old male, a college student, was referred for subjective distress. He remembered

developing facial tics at a very early age, and it had been exacerbated by the death of his grandmother a few months prior to consultation.

He felt embarrassed as he would compulsively nod his head, gesticulate with his upper limbs and make grunting noises. Due to his symptoms he felt inadequate and socially isolated himself. By 19 years old he had very frequent grimacing, echolalia and stamping of the feet. He had sought the help of innumerable physicians and traditional healers without any remission of symptoms. During the time of consultation, he was depressed, had very poor concentration and a very low self-esteem.

TREATMENT BY MASS NEGATIVE PRACTICE

(a) Theoretical Models

Cases 1 and 2 were initially treated by Behaviour Modification, utilizing the technique of mass negative practice. The method of treatment (Yates, 1970) was derived from both the Hullian and Skinnerian models, utilizing the reactive inhibition and the drive-reduction theory. This theory states that reactive inhibition would be generated as mass negative practice continues and the dissipation of reactive inhibition would be reinforcing, thereby effecting a reduction in tics. Yates (1958) conceptualized the tic as a drive-reducing conditioned-avoidance response originally evoked in a highly-traumatic situation. In such a situation, intense fear may be aroused but direct escape from the situation may be impossible. Hence, a secondary fear-response which may reduce anxiety, is in the performance of a tic.

He also experimentally demonstrated that mass negative practice resulted in a significant decline in the frequency of tics and reactive inhibition built up rapidly. When reactive inhibition reached a critical point, the patient would be forced to 'rest' and not perform the tic. The basic theory is that what will be reinforced during the rest period will be the response of not being able to perform the tic.

(b) Treatment of Cases

Walton (1961, 1964) treated two cases of multiple tics by prolonged periods of mass negative practice with a significant reduction in the frequency of tics. Lazarus (1960) also successfully treated a similar case by the same technique. In 1966, Clark reported the successful application of mass practice to compulsive obscene utterances of Gilles de la Tourette's Syndrome. Two of his three cases showed a remission of symptoms over a four-year period.

(c) Method

(i) *Case 1*

Case 1 was admitted and the frequency of his tics was recorded for the first few days of admission with a self-counting counter. He averaged 650 tics a day. As a means of feedback to progress, he was taught to count his own tics and to plot them daily on graph paper.

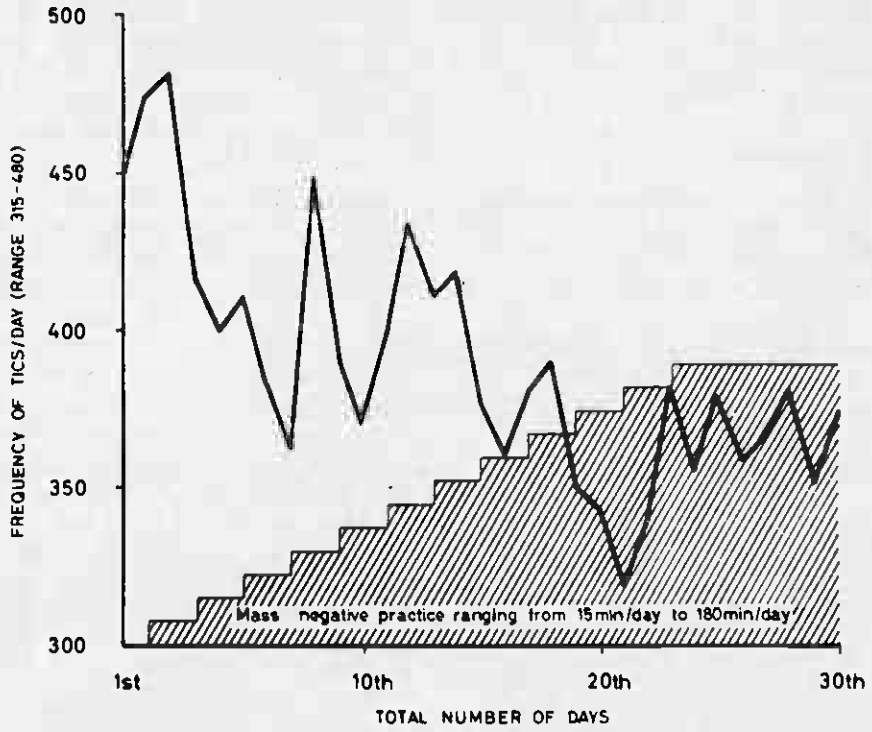
Mass negative practice was started on the third day, with initial sessions of five minutes per session, three times a day. During these sessions he was placed in a closed room and encouraged to evoke the exact tics, utterances and gesticulations which he suffered from. The duration of the mass practice was increased by five minutes per session every second day until he reached a maximum period of one hour per session, three times a day i.e. three hours mass practice per day. The sessions were kept up daily for three months and the frequency of tics was recorded daily. While the frequency of tics was reduced in the succeeding hours after each mass practice, the total frequency of tics per day was not reduced at all (see Fig. 1). However, prolonged mass negative practice did reduce the quality of tics, i.e. major tics were reduced to minor tics, although the frequency remained unchanged. Towards the end of the sessions, he was more able to keep still, without corprolalia, echolalia, feet stamping, but minor facial twitches, grimaces and hand movements remained. (He was eventually placed on Haloperidol therapy which dramatically removed the minor tics completely).

(ii) *Case 2*

Case 2 was treated utilizing the same regime of mass negative practice as with Case I. However, he managed to stay in the ward for only one month, until his corprolalia which was directed at the nursing staff, caused much furore among the nursing staff, and the author was obliged to discharge him. The results of mass negative practice (see Fig. 2) caused a reduction in the quality of tics i.e. from major to minor tics, but did not significantly reduce the frequency of tics. Although at discharge, he was less fidgety, facial grimaces, echolalia and corprolalia persisted. (He was subsequently readmitted for Haloperidol therapy which dramatically reduced his symptoms).

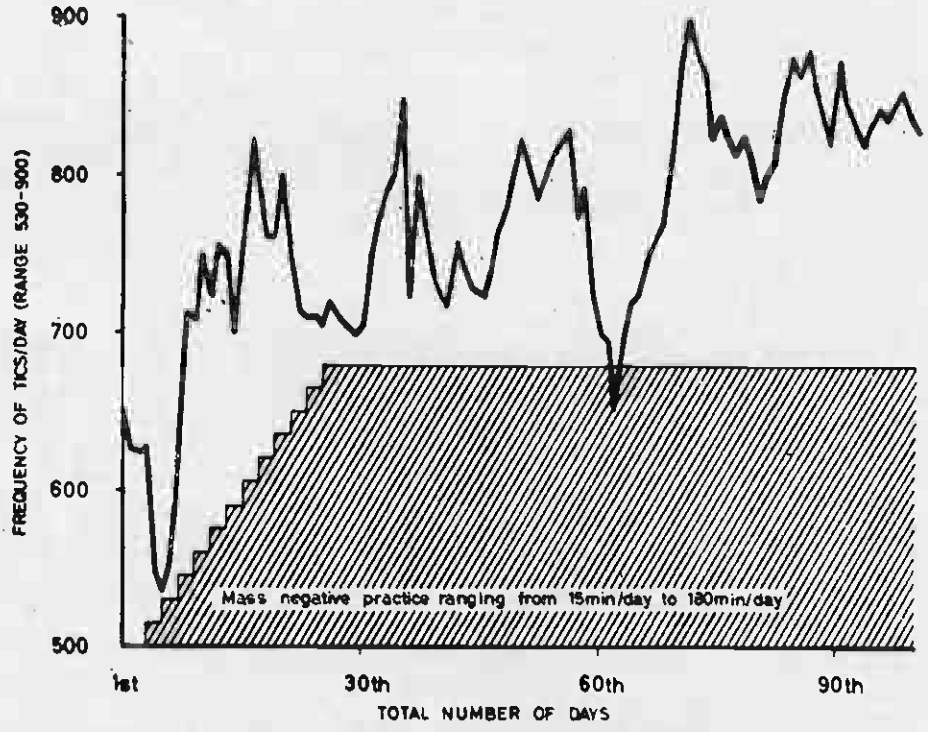
TREATMENT WITH HALOPERIDOL

A large number of psychotropic agents have been administered to such patients with discouraging results. In 1961, Seignot reported using Haloperidol in a case of Tourette's disease with dramatic symptomatic improvement. Its effectiveness



Graph 1: Case 1: GILLES DE LA TOURETTE'S SYNDROME.
FREQUENCY OF TICS RELATED TO MASS NEGATIVE PRACTICE

Fig. 1.



Graph 2: Case 2: GILLES DE LA TOURETTE'S SYNDROME.
FREQUENCY OF TICS RELATED TO MASS NEGATIVE PRACTICE

Fig. 2.

was further proven by others (Challas and Brauer, 1963, Chapel *et al*, 1964 and Shapiro, 1964). However, whenever Haloperidol was discontinued relapses occurred.

(a) Method of Treatment

Six cases of Gilles de la Tourette's Syndrome were treated with Haloperidol. (Cases 1 and 2 were subsequently placed on Haloperidol therapy after partial remissions with mass negative practice). The tics of Cases 2, 3, 4, and 5 were recorded on video-tapes for 10-15 minutes prior to treatment. (Case 6 was not video-taped as his symptoms were mild and improvement could not be significantly measured).

The video-tape recording was utilized as a controlled measurement of symptomatic improvement. The patients (Cases 2-5) were then treated with varying doses of Haloperidol at gradually increasing doses until symptomatic improvement was observed. The doses of Haloperidol ranged from 6 mg. to 33 mg. per day (see Table II). The increasing dosage was stopped when side-effects of the drug interfered with the patient's functioning. The side-effects of Haloperidol were drowsiness, parkinsonism, poor concentration, visual disturbances and sphincteric spasm leading to delayed initiation of micturation. Parkinsonism was controlled with Benzhexol, dosage ranging from 12 mg. to 36 mg. per day. Approximately one month later, they were subsequently video-taped and the degree of improvement was measured.

(b) Video-tape Technique of Recording

The video-tape recording system provided a continuous and a permanent recording of the frequency and the quality of tics in each case. Each video-tape recording was done in a similar studio condition. The patients were given exact instructions to behave naturally in a sitting position. The studio environment remained constant for each recording.

Standard criteria for coding observation of tics in different body components were recorded simultaneously by two independent observers every fifteenth second before and after treatment with Haloperidol. The raw data was collected from the video-tape recordings of the first to the 3½ minute (i.e. total observation period of 2½ minutes) and of the 6th minute to the 8½ minute (i.e. 2½ minutes observation) before and after treatment with Haloperidol. The reliability of different observers' scores was calculated by a simple percentage agreement-disagreement of raters' scores (see Table III). The range of percentage agreement before treatment was 70·8% to 94·3% and after treatment was 94·8% to 100%.

(c) Results of the Study

Degree of Percentage Improvement following Haloperidol Therapy

The total mean scores of all four cases for each body component of movements before and after treatment with Haloperidol were noted. (See Table II).

There was a marked reduction in scores for each body component of movement after treatment. The overall percentage improvement of tics after treatment with Haloperidol ranged from 52·5% (in Case 4) to 98·5% (in Case 3). There was a remarkable improvement in terms of reduc-

TABLE II

MEAN SCORES OF DIFFERENT BODY COMPONENTS OF MOVEMENT IN CASES 2-5 OF GILLES DE LA TOURETTE'S SYNDROME BEFORE AND AFTER TREATMENT WITH HALOPERIDOL

	Head Movement		Grimacing		Upper Limbs		Lower Limbs		Trunk	
	Bf. Tr.	Af. Tr.	Bf. Tr.	Af. Tr.	Bf .Tr.	Af. Tr.	Bf. Tr.	Af. Tr.	Bf. Tr.	Af. Tr.
Case 2	29	5	32	3	52	8	56	10	33	4
Case 3	35	5	26	12	43	7	28	13	9	1
Case 4	9	0	13	1	49	0	47	0	26	0
Case 5	24	16	14	4	29	25	61	22	10	2
Mean Scores	24·0	6·5	21·3	5·0	43·3	10·0	48·0	11·3	19·5	1·8

Overall mean score before treatment = 156·1 Overall mean score after treatment = 34·6

TABLE III

PERCENTAGE IMPROVEMENT IN VARIOUS BODY COMPONENTS OF TICS IN CASES 2-5 OF GILLES DE LA TOURETTE'S SYNDROME FOLLOWING TREATMENT WITH HALOPERIDOL

	Head Mov. (%)	Grimacing (%)	Upper Limbs Mov. (%)	Lower Limbs Mov. (%)	Trunk Mov. (%)	Overall Improv. (%)
Case 2	62·8	96·7	84·6	82·1	87·9	86·8%
Case 3	88·9	53·9	83·7	53·6	88·7	73·8%
Case 4	100·0	92·3	100·0	100·0	100·0	98·5%
Case 5	33·3	71·4	13·8	64·0	80·0	52·5%

tion of tics in all components of the body following treatment with Haloperidol (See Table III).

Case 6, which was not video-tape recorded, also showed marked clinical remission of symptoms following Haloperidol therapy.

CONCLUSION

Two cases of Gilles de la Tourette's Syndrome were initially treated with prolonged periods of mass negative practice. Results indicated that mass negative practice did not significantly reduce the frequency of tics, but did reduce the quality of tics. The results were contrary to that of Clark (1966) where he obtained a four year remission of symptoms with two cases of Tourette's Syndrome treated by a similar technique.

Six cases of Gilles de la Tourette's Syndrome were subsequently treated with Haloperidol and the degree of improvement (in four cases) was measured by the technique of video-tape recording. The overall percentage improvement following Haloperidol therapy was dramatic in terms of reducing the quality and the frequency of tics.

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