apply to patients who share a fixed group of clinical phenomena, but no published studies have demonstrated other markers of the existence of a disease entity, such as genetic linkage, predictable response to a particular treatment, or the sharing of a common prognosis.

Thus a syndrome first described by Hippocrates in the context of a quite different conceptualisation of mental illness has perhaps outlived its usefulness. We would subscribe to the view expressed by Lehman (1980) that "it would be advisable not to perpetuate the existence of this questionable syndrome in the literature".

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The Pisa Syndrome: A Report of Two Cases

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Ekbom *et al* (1972) described dystonic syndromes that appeared as a side-effect to treatment with the butyrophenone group of neuroleptic drugs. These symptoms consisted of tonic flexion of the trunk to one

side, accompanied by its slight rotation, in the absence of other concomitant dystonic symptoms. They called this the 'Pisa syndrome'. A literature survey of neuroleptic-included extrapyramidal side-effects gave little reference to this syndrome (Sovner & DiMascio, 1978; Berger & Rexworth, 1980), but two cases that fit the description of this side-effect are presented here.

Case 1

A 57 year old man was admitted to the Douglas Hospital in 1975 with a diagnosis of general paresis of the insane; this was confirmed by positive serological testing in blood and cerebro-spinal fluid. Due to accompanying behavioural problems, as well as a chronic delusional system, he was treated with chlorpromazine, the highest daily dose being 1600 mg in 1979 and the lowest 600 mg. In November 1980, and while receiving 900 mg chlorpromazine per day, the patient was noted to develop tardive dyskinesia of a bucco-lingual nature, with frequent tongue movements. This was noticed after a sudden reduction of chlorpromazine from 1600 mg to 900 mg, and has persisted till the time of writing. In March 1982, while receiving 1200 mg of chlorpromazine per day, the patient was noted to have severe drooling; this was treated with benztropine, 4 mg daily. From September 1982 until April 13 1983, the patient received chlorpromazine, 900 mg per day. On April 12 1983, he was noted by the nurses to lean to the left. He was examined the following day by the author, who found the patient to be flexed to the left, and while standing, his trunk was bent to the left and slightly rotated backwards in the same direction. A neurological examination showed no evidence of stiffness in either both upper or lower limbs, and the reflexes were normal on both sides. Chlorpromazine was decreased to 600 mg per day and benztropine 4 mg per day added once more. The patient was examined on a daily basis and on April 19, he was still bending to one side. This side-effect continued until April 26, without any change in severity; then all medication was discontinued, and the patient's symptoms disappeared within 16 hours. However, his tardive dyskinesia worsened, and his tongue movements were now more frequent, being rated as moderate. On June 28, he was restarted on methotrimeprazine 100 mg four times a day because of aggressive behavior and impulsive acts; he had only received two doses when he was noted to be leaning to the left. Neuroleptics were discontinued, and the patient's condition improved to the extent that, when examined the following day, he was symptom-free.

Case 2

A 32 year-old male was first diagnosed as schizophrenic in 1972, and had since had several admissions to our hospital, all with the same diagnosis. He received fluphenazine enanthate, 50 mg IM every two weeks since 1975, but was admitted in May 1983 in an acute paranoid state. Chlorpromazine, 800 mg per day was added to the fluphenazine, which was increased to 1200 mg per day on the first of June. Seven days later, he was noted to lean to the right; on examination the same findings as described above were noted. His trunk was flexed to the right, and no evidence was found of stiffness in either upper or lower limbs, nor any tardive dyskinesia. The patient was given benztropine, 4 mg per day, in addition to his regular medication, with little effect; two injections of benztropine were given on June 10 and June 13 because of an aggravation of his postural disturbance. However, on June 13, he was found dead in bed; an autopsy was performed but no report has been made available.

Discussion

The two cases presented here fit the description by Ekbom *et al* (1972) of the 'Pisa syndrome', having shown the peculiar postural disturbance described, i.e. flexion of the trunk to one side, with slight rotation backwards. The symptoms were constant during the patients' activities, as well as while sitting or standing. No other dystonic manifestations were noted at the time of the appearance of these symptoms i.e. no oculogyric crises, torticollis or tonic spasms of the jaw muscles. As in Ekbom's case 2 (1972), an elderly woman who exhibited tardive dyskinesia before the development of the Pisa syndrome, the first of our two patients showed evidence of tardive dyskinesia that had its onset well before the appearance of the Pisa syndrome and that became worse after discontinuing neuroleptics. However, the manifestations of the Pisa syndrome dramatically improved after the drugs were withheld.

This syndrome has occurred in patients receiving antipsychotic medications for prolonged periods and may be considered as a manifestation of tardive dystonia (Burke *et al*, 1982 a, b). This is to be differentiated from the acute dystonia that usually occurs in the early stages of neuroleptic treatment (Marsden *et al*, 1975; Sovner & DiMascio, 1978; Marsden & Jenner, 1980).

The coexistence of tardive dyskinesia and dystonic symptoms (as seen in patient 1), has been described in Ekbom's *et al's* case 2, while Nasrallah *et al* (1980) and Gardos (1981) each described a case where tardive dyskinesia had been present before the appearance of dystonic symptoms. Munetz (1980), however, described dystonic symptoms developing in a male patient who subsequently also had tardive dyskinesia. This combination of dystonic symptoms and tardive dyskinesia can pose problems of differential diagnosis. Tardive dystonia is similar to tardive dyskinesia in that patients may suffer from the disorder for prolonged periods and that both may be refractory to treatment (Burke *et al*, 1982). Sometimes, the differentiation between the two conditions may be impossible, especially when the oro-mandibular region is affected (Marsden, 1976). Tardive dystonia may be aetiologically and pathophysiologically related to tardive dyskinesia, although anticholinergic medication may benefit some patients with tardive dystonia (BTP,

1983), while they typically worsen the abnormal movements of tardive dyskinesia. Acute dystonias, on the other hand, should be differentiated from tardive dystonia by the time course relative to drug exposure (Mackay, 1982) and by the response to treatment (Flaherty & Lahmeyer, 1978; Mann *et al*, 1979; Menuch, 1981; Moss & Grien, 1982).

The treatment of tardive dystonia is not well established (Burke *et al*, 1982b; Kwentus *et al*, 1984). Anticholinergic medication may benefit some patients, but not others. Tetrabenazine, carbamazepine, propranolol, and clonazepam have been reported to benefit some patients (Burke *et al*, 1982a). In one patient, electroconvulsive therapy led to a dramatic disappearance of the symptoms (Kwentus *et al*, 1984). In our patients, the symptoms disappeared with antipsychotic drug withdrawal, while anticholinergic medication did not affect the symptom's manifestations.

Thus, clinicians should be alert to the possibility of the Pisa syndrome and other tardive dystonias developing in patients on long-term antipsychotic medication, and should not consider all trunk and limb movements in such patients as tardive dyskinesia.

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