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Review Article

Dystonia – A Potential Psychiatric Pitfall

D. G. CUNNINGHAM OWENS

The manifold presentations of dystonia have been acknowledged as a spectrum of illness. Dystonia is primarily idiopathic or drug-induced; purely psychogenic causes would seem to be rare, although such an aetiology is commonly assumed. Response to medication in idiopathic cases is unpredictable, although anticholinergic medication and behavioural programmes are being evaluated.

It may be true to say that at present general psychiatrists come into contact with disorders of movement as often as general neurologists. Despite a great deal of interest in the subject of movement disorders in the psychiatric literature in recent years, there is evidence that in routine clinical practice a considerable amount of abnormality of this type is overlooked (Weiden *et al*, 1987). Psychiatrists still seem unattuned to seeking out movement disorder, especially if mild, and perhaps equally important, may feel themselves ill-equipped to categorise such essentially neurological abnormality. In view of the prevalence and potential significance of these features, oversight in this area, whatever its basis, represents a major limitation to comprehensive patient assessment.

While hyperkinetic disorder of choreoathetoid type has been extensively described in the psychiatric literature, relatively little has been written about dystonia, a concept which has itself undergone refinement by neurologists in recent years.

History

The term 'dystonia' has a varied history. Some authors applied it in what might be called a generic sense. Thus, for Wilson (1940) dystonia meant "any variability in muscle tone," while Denny-Brown (1962) emphasised the "fixed abnormalities of posture" that result from persistent tonal changes, as is to be found for example in hemiplegia or Parkinsonism. This 'generic' usage has largely been abandoned, and dystonia is now usually reserved for either a specific syndrome or, increasingly, for the abnormalities characteristic of this syndrome.

Hammond (1871) was the first to describe movements having the major characteristics of this type of disorder – namely gross muscle spasms and resulting distortions of normal posture. In his two patients the limbs were affected resulting in an *inability to hold a fixed position*, a sign he referred to as 'athetosis'. Athetosis and dystonic abnormality differ only in distribution but not essentially in quality. Thus 'athetosis' is strictly 'distal dystonia' (Marsden, 1976*a*) – or, confusingly, dystonia has been viewed, in Wilson's (1940) terminology, as 'proximal athetosis'!

Schwalbe (1908), in his doctoral thesis, described a syndrome of 'chronic cramp' in three siblings of a Lithuanian Jewish family which, being a psychiatrist, he attributed to psychological (i.e. hysterical) causation. Three years later, however, the neurologist Oppenheim described six new cases and asserted that the basis of the disorder was not psychological but a variability of muscle tone with consequent distortion of normal posture. To reflect this he suggested the name by which the condition is still known, 'dystonia musculorum deformans' (Oppenheim, 1911). Since then neurologists have frequently used the term 'dystonia' as a sort of short-hand for this uncommon, often familial disorder. Other early workers on the subject chose to emphasise that the characteristic movements of this condition had a sustained and typical twisting quality (Ziehen, 1911; Flatau & Sterling, 1911), a view accommodated in the alternative name of 'torsion spasm', also still widely used.

Dystonia musculorum deformans (DMD) is a rare disorder. It has however been recognised for many years that involuntary movements with the same sustained, twisting characteristics as those typical of DMD could affect isolated muscle groups or local body parts. These abnormalities tend to have a later age of onset and rarely generalise to involve the whole body. The names traditionally applied to these conditions reflect their local distributions, such as blepharospasm, torticollis, or writers' cramp. In addition to their descriptions in neurological texts, these conditions have, interestingly, also figured heavily in the pages of psychiatric textbooks (e.g.

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Abse, 1966) as examples of conversion hysteria, illustrating the parallel but separate paths the two specialities have often taken in the past century.

In the last few years there has been a reappraisal of the position of localised dyskinesias of this type in relation to the more generalised form of disorder. This has resulted in a unifying classificatory system encompassing all the dystonias.

Definition

In a condition with such a varied history, producing a unified definition is not easy. Early on Wechsler & Brock (1922) emphasised two main clinical forms which they referred to as 'myostatic' and 'kinetic'. Since then some authors have emphasised the static elements of the disorder in terms of the attitudes produced as a result of the abnormal muscle contractions, while for others the dynamic aspect of the abnormal contractions themselves was more important (for review of definitions see Fahn *et al* (1987)).

In 1984, an *ad hoc* committee of members of the Scientific Advisory Board of the Dystonia Medical Research Foundation produced the following definition:

> "Dystonia is a syndrome dominated by sustained muscle contractions, frequently causing twisting and repetitive movements, or abnormal postures." (Fahn *et al*, 1987)

Classification

Dystonias have been classified by age of onset, by cause, or by distribution (Table I). Classification by age of onset is valuable because of the prognostic implications. Age of onset is probably the most important factor with regard to severity and the likelihood of generalisation (Marsden *et al*, 1976; Cooper *et al*, 1976; Fahn, 1986). Thus, the younger the onset (especially under ten years of age) the more likely is generalisation to take place. By contrast generalisation is rare with onset after 40 years (Marsden *et al*, 1976; Fahn *et al*, 1987). Psychiatrists are most likely to come in contact with adult-onset cases, so classification by cause or distribution is more useful to their practice.

TABLE I
Proposed classification of dystonia

Classification by:	*Criteria*
Age of onset	<12 years of age
	13–20 years of age
	>20 years of age
Aetiology	Idiopathic
	(a) sporadic
	(b) familial
	Symptomatic
Clinical distribution	Focal
	Segmental
	Multifocal
	Generalised
	Hemidystonia

After Fahn *et al* (1987).

While most dystonias are idiopathic, symptomatic cases associated with neuroleptic drug treatment are those most frequently encountered in psychiatric patients. It is encumbent on all clinicians however not to be blinkered in their approach to these abnormalities as many other causes of symptomatic dystonia have been reported apart from neuroleptic drug exposure. The reader is referred to the exhaustive differential diagnosis presented by Fahn *et al* (1987) for details, where over 40 associated disorders are listed. Although most of these are clearly neurological (and some very rare), a number may be seen in psychiatric contexts. These include Wilson's disease, Huntington's disease, Lesch–Nyhan syndrome, encephalitis, head injury, intracranial tumours, multiple sclerosis, cerebrovascular accident, carbon monoxide poisoning, and exposure to centrally acting drugs other than dopamine-blocking agents (see below).

A descriptive classification based on distribution is clinically the most straightforward. This divides dystonia into five main types:

(a) focal: affecting one body area only
(b) segmental: affecting two or more contiguous body areas
(c) multifocal: affecting two or more non-contiguous body areas
(d) generalised: affecting multiple, disparate body areas
(e) hemidystonia: affecting ipsilateral limbs, with or without involvement of ipselateral trunk or face.

By this classification a number of differently named clinical entities are subsumed under recognisably similar terminology. The arguments justifying this have been presented by Marsden (1976*a*) and Fahn *et al* (1987). Thus blepharospasm, torticollis, and writers' cramp all become examples of *focal* dystonia, while Meige's syndrome (see below) becomes an example of a *segmental* dystonia.

Hemidystonia is usually symptomatic (Fahn *et al*, 1987), secondary to a contralateral brain lesion, especially of caudate/lenticular nucleus/thalamus (Marsden *et al*, 1985). It must be remembered however that movement disorder associated with

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neuroleptic exposure can present unilaterally (Waziri, 1980; Myslobodsky *et al*, 1984; Wilson *et al*, 1984), although usually the abnormality is then restricted to one limb.

General clinical characteristics

Dystonia has two components: the active or 'kinetic' movements themselves (the dyskinesias), and the maintained postural abnormalities or persistent attitudes resulting from abnormal muscular contraction. These may occur singly or in combination. Any striated muscle can be involved, but in the non-generalised varieties, the head and neck are most commonly affected (craniocervical dystonia). Likewise, when drugs and other potentially causative factors can be ruled out, dystonia is probably the most common form of *spontaneous* craniocervical dyskinesia (Jankovic, 1988*a*).

Onset may be rapid, but usually the condition evolves slowly, over months or years. The initial complaints may be vague and non-specific, such as nuchal ache or eye irritation, or muscle tension may be evident only when the patient performs some voluntary activity in the affected area ('action dystonia'). With progression, activity in distant muscle groups may precipitate dystonia ('overflow' phenomenon). As a general rule, dystonic movements are invariably aggravated by voluntary activity, although eventually movements and abnormal postures may be evident at rest.

It has recently been appreciated that the movements of dystonia can vary greatly in the speed with which they develop. While typically they tend to be slow and sinuous, some can be rapid in onset and difficult to distinguish from other dyskinesias such as tics or chorea, and may even have the 'lightening' quality of onset of myoclonus – so-called myoclonic dystonia (Obeso *et al*, 1983). Like their slower counterparts, these rapid movements have the same twisting characteristics and tend to be continual and repetitive. It has been suggested in addition that abnormal movements should be sustained for at least one second at the point of maximum contraction to qualify as dystonic (Fahn, 1984). Other involuntary movements may however occur at the same time, a particular association being noted with postural tremor, and a family history of essential tremor may also be found (Marsden, 1976*a*).

Dystonias have unusual characteristics, some of which are shared with other extrapyramidal movement disorders, some of which are unique. Thus in common with other involuntary movements they are influenced by the degree of mental arousal, and are consequently exacerbated by anxiety or other adverse mental state features or by stress in general. Conversely they are eased by calm, relaxation or sedation, and may be suppressed completely during hypnotic states. Such psychological factors may underly a certain degree of periodicity in symptom expression in that movements can be evident on some days but not on others, and sustained postures can relax intermittently. Like most other dyskinetic states, dystonias disappear during sleep. Specific focal dystonias may exhibit specific precipiting factors (see below) and a wide range of task-specific dystonias are precipitated only by use of occupational implements of a certain type, size, or weight.

In addition, however, dystonias have certain other characteristics that are frankly bizarre. The motor action precipitating the dystonia may be highly selective, even within a specific body region, preserving other actions. Thus a patient with deforming dystonia of the legs which precludes normal forward gait may be able to walk backwards or run freely. One of the author's patients with severe oromandibular dystonia and profound scoliosis lost both his disfiguring movements and his postural deformity on the dance floor, and has taken up dancing with vigour since recognising himself the association with symptom suppression.

Many patients find, at least in the earlier stages of their disorder, that certain tactile or proprioceptive stimuli can reduce or even abolish their dystonia. These various activities 'devised' by the patient have been referred to as 'sensory tricks' or 'antagonistic gestures', and are said to be unique to dystonia and hence of diagnostic value (Fahn *et al*, 1987). Examples of this are attenuation of head deflection in torticollis by touching the chin, or holding the back of the neck with the opposite hand to the direction of rotation. One patient found that while this latter procedure did not markedly alter the degree of displacement from her severe torticollis, it did greatly ease the subjective discomfort. Another patient, in effect functionally blind with blepharospasm, could achieve a sufficient degree of eye opening to allow walking by rubbing his upper dental margin.

In view of these unusual characteristics it is easy to see how dystonias could be considered primarily psychogenic disorders.

Major clinical types

Acute dystonias

Acute dystonias are probably the commonest (*Lancet*, 1985). Although secondary to antidopaminergic medication, they are best classified separately from

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other symptomatic dystonias in view of their sudden onset and brisk response to treatment, which implies a distinct, or possibly more transient, pathophysiology.

It is important to remember that any drug with dopamine-blocking potential can cause these bizarre abnormalities. Thus while all classes of typical neuroleptics are undoubtedly the commonest causative agents, other drugs less frequently encountered in a psychiatric setting may be implicated, such as the antiemetic substituted benzamide metaclopramide and certain antihistamines. One recent immigrant to the UK from West Africa was referred to the author as a hysterical malcontent expressing his contempt for Western culture by permanently extruding his tongue. As a poor traveller he had taken travel-sickness pills for the flight and his 'contempt' was instantly accommodated following intravenous anticholinergic.

The distribution of acute dystonic reactions interestingly parallels that of the idiopathic dystonias in that it depends to some extent on age. Thus in children they are often severe and generalised, whereas in adults they are more usually (though not exclusively) craniocervical (Ayd, 1961; Marsden *et al*, 1975). They develop suddenly, usually within 24–48 hours of first exposure to the provoking agent, or after dose increment (Ayd, 1961), although may be delayed until 72 hours or longer with depot preparations (Johnson, 1973). They can last, untreated, from minutes to hours, and may come in runs, with periods of relative quiescence between.

The clinical features are varied and often bizarre, and can include aversive eye movements, blepharospasm, trismus, mouth opening, tongue protrusion, grimacing, distortions of lips, displacement/rotation of head and neck, hyperpronation, finger/wrist posturing, opisthotonos and other axial distortions, and gait disturbance. Dysarthria, dysphagia, jaw dislocation, tongue amputation, and respiratory stridor with cyanosis may ensue. A promonitory phase of local cramp-like discomfort or tightness of jaw or tongue may occur, making chewing, swallowing or speaking difficult, or such features may be the sole manifestation or disorder. It is crucial to view all 'lists' of acute dystonic symptoms with circumspection as none can be comprehensive in recording all the possible combinations that will be encountered clinically.

Acute dystonias with neuroleptic usage may be more frequent than is often assumed. Ayd's (1961) figure of 2.3% is still quoted, but his method of hierarchical classification of extrapyramidal reactions means that this figure refers to those of his sample who experienced only acute dystonia without subsequent akathisia or Parkinsonism. Addonizio & Alexopoulos (1988) suggested a prevalence of 25% overall; in addition they demonstrated the clear effect of age on prevalence – while 2% of elderly patients (mean age 69 years) on neuroleptics had shown dystonic reactions, the comparable figure for young patients (mean age 26 years) was 31%, a finding not attributable to differing prescribing habits. While it is generally believed that high-potency antipsychotics are most likely to be implicated, Addonizio & Alexopoulos also found that in terms of chlorpromazine equivalents, young patients who developed acute dystonias were receiving *lower* doses than those who did not, implying an element of individual susceptibility. One factor in this may be gender, as males appear to be twice as commonly affected as females (Swett, 1975; Ayd, 1961, 1983).

Acute dystonias, in addition to the frequently grotesque appearances they impart, may be painful and invariably are exceedingly distressing. Following the introduction of neuroleptics they were often misdiagnosed as tetanus (Melvin, 1962), status epilepticus, or manifestations of catastrophic CNS infection, resulting in needless investigations such as lumbar puncture and inappropriate interventions such as tracheostomy (Ayd, 1961). In the author's experience, however, they are still sometimes misdiagnosed as 'hysterical'. There is little excuse for such an error in psychiatric practitioners. Any patients with the sudden onset of subjective stiffness or typical movement and postural disorder in any body part, *especially* if the clinical appearance is bizarre, should be given a trial of parenteral treatment, whether or not an appropriate drug history can be obtained. Patients may not wish to be forthcoming on such matters, may be ignorant of what they have ingested, or may be so distressed and disabled, especially by lingual or oropharyngeal/laryngeal dystonia, that they are unable to respond to specific questions adequately.

A trial of treatment should not await resolution of the drug question. Appropriate treatments are safe, readily available and rapidly acting (within minutes). Anticholinergics are now the most widely used. Notwithstanding the above, however, the euphorogenic properties of these preparations (Shader & Greenblatt, 1971; Jellinek, 1977; Kaminer *et al*, 1982) and the pharmacological sophistication of many psychiatric patients must be borne in mind when faced with repeated requests for anticholinergics at inappropriate phases of neuroleptic exposure or in the continuing absence of objective signs. In the past barbiturates, diazepam, and the antihistamine diphenhydramine have also been reported as effective but have largely been superseded.

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Oculogyric spasms

J. P. Albrecht gave the first reliable account of ocular spasms occurring in epidemic encephalitis as far back as 1695 (quoted in Jelliffe, 1929). From 1921 onwards, so-called 'oculogyric spasms' were increasingly described as a late sequela of encephalitis lethargica, although a similar phenomenon was also noted in neurosyphilis. Episodes occurred almost exclusively in the context of the Parkinsonism that followed the acute illness (Jelliffe, 1929; Wilson, 1940) and were characterised by the sudden deviation of both eyes upwards or upwards and to one side, usually the right. Spasms were maintained ('tonic') or less commonly jerky ('clonic') and were often associated with head/neck deviation ('cephalogyric spasms', usually retrocollis). They lasted from minutes to hours. Pain was sometimes noted but a particularly unnerving anxiety or fearfulness was regularly experienced (Jelliffe, 1929; Wilson, 1940). Precipitating factors included any states of extreme emotion as well as a range of physical factors such as excessive sensory stimulation (e.g. bright light, noise, heat) or fatigue and exercise (Van Bogaert, 1934). Sometimes the patient recognised 'warnings' (Wilson, 1940). A diurnal variation with increased afternoon/evening frequency was often noted, possibly related to the effects of increasing muscular fatigue.

Present-day psychiatrists would recognise some of the above as comparable with what is seen with antidopaminergic treatment. Usually this occurs as part of an acute reaction following initial exposure but the author has encountered such spasms as an isolated phenomenon usually in the course of gradual reduction of depot regimes.

The interest in this shown by a previous generation of neurologists has not been shared by present-day psychiatrists and nothing is now known of precipitating factors, 'warnings', diurnal variation, or the prevalence of associated abnormalities. This is of interest as it is clear that post-encephalitic ocular deviation was but one manifestation of an often complex neuropsychiatric disturbance (Jelliffe, 1929). Dystonia in other sites, autonomic overactivity, altered consciousness, visual misperceptions and compulsive 'forced' thinking/ruminations were all described (Jelliffe, 1929; Wilson, 1940), as were speech disturbances either premonitory to or during an attack. These included palilalia, stereotyped phrasing, echolalia, logorrhoea, mutism, and forced shouting (so-called 'klazomania') (Van Bogaert, 1934; Wohlfart *et al*, 1961). The author is unaware of work addressing all or any of these points in the context of drug causation.

When a range of other neuropsychiatric features or prominent affective disturbance is present the condition would justify being referred to as a 'crisis', but with uncomplicated eye deviation 'oculogyric spasm' or simple 'oculogyrus' would seem more appropriate.

Focal dystonias

Torticollis

Although uncommon, idiopathic spasmodic torticollis is the most prevalent of the focal dystonias (Duane, 1988). Formerly it was estimated to have accounted for 1 in 1200 psychiatric referrals (quoted by Lees, 1985). The maximum age of onset is in the fifth decade and while some authors report an equal sex prevalence (Lees, 1985), others note a female excess (Duane, 1988).

Although the sternocleidomastoid is the most commonly affected muscle, there is no single characteristic presentation. Contrary to general belief, the pattern of muscular involvement is often highly complex, which results in postural distortions encompassing every possible head position, not just the simple rotational abnormality implied in the name (Fahn, 1984). It has been pointed out that only by marked effort can normal individuals simulate these changes (Lees, 1985).

Abnormal head position is noticed first by others in about 25% of cases, and in a minority may be preceded by nuchal discomfort. Movement may initially be triggered by bimanual tasks such as driving, or by attempts to maintain a formal posture, as for example in a public gathering. Deviation can be sustained or intermittent, with progression in severity over approximately 18 months. Remissions, especially in those under 40, are not uncommon (approximately 20%), but neither is subsequent relapse (Duane, 1988).

Pain, especially at the points of insertion of the sternomastoid and upper trapezius, is common, although later secondary degenerative joint disease may be a greater source of discomfort than muscle pain.

'Sensory tricks', including touching the chin, vertex, or back of the neck with the hand contralateral to the direction of rotation, may alleviate symptoms, as may yawning, swimming, or lying down. Indeed, patients may report themselves free of dystonia for up to an hour after rising in the morning. Emotional distress, reading, writing, menstruation, pregnancy, and alcohol (especially 'the morning after') have all been reported as exacerbating influences (Duane, 1988).

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In addition to head displacement, examination may reveal scoliosis (50%), palpable muscle spasms and even enlargement, other facial dystonias, and head tremor, especially of horizontal or 'negation' type.

The aetiology is in general unknown. A small number of cases appear to be familial (Gilbert, 1977) and associations have been reported with head/neck trauma (Sheehy & Marsden, 1980; Duane, 1988), diastolic hypertension, thyroid disease, especially in women, and alcoholism, especially in men (Duane, 1988). There are no predisposing personality attributes (Cockburn, 1971) and depression when present is a secondary rather than an aetiological factor (Tibbetts, 1971).

Less common variants are laterocollis (head to shoulder) and the bilateral spasms resulting in retrocollis (10% of idiopathic cases) and antero- or antecollis (5%).

Blepharospasm

Blepharospasm is characterised by intermittent or persistent involuntary closure of the eyes due to spasmodic bilateral contractions of the orbicularis oculi muscles. It usually begins in the sixth decade and there is a female preponderance (Jankovic *et al*, 1982). In the early stages it may present with non-specific discomfort or a complaint of excessive, uncontrollable blinking (blepharoclonus), and may be unilateral. It is often misdiagnosed as 'dry eyes' (Jankovic, 1988*b*). Progression leads to increasing incapacity and ultimately functional blindness.

In the early stages precipitating factors include upward or downward gaze, bright light (patients often wear dark glasses), strong direct wind, smoke and other air pollutants, fatigue, and emotional distress. Active concentration may on the other hand sustain eye opening, leading to a false impression of the degree of disability at examination (Marsden, 1986).

A wide range of 'tricks' may help, such as touching the forehead, pinching the neck, humming, or singing (Jankovic, 1988*b*). Severity may fluctuate.

In addition to forced eye closure, another clinical variant is mild to moderate tonic contraction of orbicularis oculi that results in persistent narrowing of the palpebral fissure (Tolosa & Marti, 1988). This type rarely interferes with vision but imparts a rather intimidating look to the patient. The author has seen several cases of this in patients on long-term neuroleptics where the somewhat menacing expression had been attributed to the mental state. This may be a commoner abnormality in such patients than has hitherto been appreciated.

Occasionally, once the eyes have been closed by involuntary contraction, the patient experiences difficulty in opening them again even though there is no involuntary activity opposing their efforts. This has been referred to as 'apraxia of eye-lid opening' (Goldstein & Cogan, 1965) or, perhaps more accurately, 'involuntary levator inhibition' (Lepore & Duvoisin, 1985), and has been reported as a common finding in Meige's syndrome (Tolosa & Marti, 1988).

Isolated blepharospasm is often referred to as 'essential' in type, but 80% of cases with eye signs show additional dystonic abnormality in eyebrow, facial, oromandibular, lingual, laryngopharyngeal or cervical muscles (Jankovic, 1988*b*), so the term 'essential blespharospasm' should be used with care.

Excluding local ophthalmological disorders, the cause of blepharospasm is unknown, although in psychiatric practice this is overwhelmingly related to neuroleptic drug exposure.

Writers' cramp and the occupational dystonias

Writers' cramp (scrivener's palsy, *schreibkrampf*, *crampe des ecrivains*) is probably one of the oldest known forms of dystonia (for a historical account see Sheehy & Marsden, 1982; Lees, 1985) and the one which has in the recent past been most strongly associated with psychogenic causation. Before the emergence of psychoanalytic theory in the 1920s and '30s, however, it was quite clearly considered to be a (central) neurological disorder. This view has recently been forcefully restated in the neurological literature.

Age of onset is rather younger than with other focal dystonias, with the maximum between 20 and 50 years. A marked male preponderance used to be described, probably reflecting the predominance of males in the clerical work force (Lees, 1985). More recent studies report an equal sex distribution (Sheehy & Marsden, 1982). The disorder does not appear to be more frequent in left-handed people (Lees, 1985).

A great deal has been made of the psychogenesis of writers' cramp (Pai, 1947; Crisp & Moldofsky, 1965). While emotional distress may be relevant as a precipitating or exacerbating factor, Sheehy & Marsden (1982) have noted how most patients are exceedingly resilient in dealing with their disability and cannot be considered psychologically exceptional. Furthermore, in their study of 29 cases, these authors used the Present State Examination to study mental state and concluded that "psychiatric factors are either accidental [i.e. no different in type or frequency from the general population] or secondary".

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Initial complaints are often vague – usually a recognition of a change in handwriting. Speed of writing may slow or quality of script deteriorate, or a difficulty in moving pen across paper may have been noted. Patients may be aware that they now grip the pen too tightly and push it clumsily through the paper or that they can no longer keep to straight lines. The hand may ache readily. It is important to note that in Sheehy & Marsden's (1982) series only 25% complained of cramps *per se*. Tremor is present in up to 50% of cases, however. (Recently focal task-specific tremor states have also been described both in isolation and in association with task-specific dystonias (Rosenbaum & Jankovic, 1988).) Such complaints are more striking when the patient has to write under pressure, such as during examinations or to meet a deadline (Liversedge, 1961). Writing even single words may become impossible.

Signs are usually striking, with clumsiness in grasping the writing implement, a tight grasp and exaggeration of the normal, semiflexed posture of the digits and hyperextension of the distal interphalangeal joint of the index finger. With writing, typical dystonic postures develop in fingers, wrists and arm, so that the fingers extend, the wrist lifts from its resting surface, the elbow elevates and the forearm pronates (Marsden, 1986). Reduction of arm swing on walking may be noted on the affected side, but other objective signs are usually lacking.

'Tricks' to help writing include exclusion of the index finger in holding the pen, switching to a thicker implement such as a felt-tip pen, using the unaffected hand to 'guide' the pen, or writing standing up. Half of Sheehy & Marsden's patients learned to write with the other hand, but a third of these developed dystonic problems on the contralateral side also. Even when writing on paper is impossible, subjects may experience no difficulty writing 'free-hand' in air or on a blackboard (Liversedge, 1961; Lees, 1985).

The problem is intermittent from the outset in only about 25% of cases. Some begin and remain as *simple* writers' cramp (i.e. the difficulty affects writing only); others show features of *dystonic* writers' cramp from the start (i.e. spasms affecting other dexterous tasks). About a third of 'simple' cases progress to the 'dystonic' form of the disorder, sometimes after many years. Although spontaneous partial remissions have been described, no patient in Sheehy & Marsden's series experienced any prolonged remission. This is therefore a disabling disorder with potentially serious employment consequences.

A great variety of other so-called occupational dystonias or 'craft palsies' have been described (Hunter (1978) for example listed 55). Some of these, because of changing work practices, are now rarely, if ever, seen – for example dystonias in telegraphists, formerly a major problem (Ferguson, 1971), and in money-counters. Others still seen include typists' dystonia, foot dystonias in ballet dancers, dystonias in musicians, especially pianists (Hochberg *et al*, 1983), and loss of dexterity in professional sportsmen whose sport involves the use of an implement, such as golfers, and darts and snooker players.

All these conditions share in common the breakdown of a particular motor skill essential for the normal exercise of the individual's occupation (Lees, 1985). In such cases, traumatic and orthopaedic causes of dysfunction must be carefully excluded.

Laryngopharyngeal dystonia

Dystonia involving laryngopharyngeal muscles is rare but can cause dysphonia and dysphagia (Marsden, 1976*a*, 1986; Lees, 1985).

The major type of laryngeal involvement is an action dystonia resulting in abductor spasm with attempted phonation (Fahn,1984). Speech is strained, hoarse, and staccato ('spastic' or 'spasmodic' dysphonia) and there may be a sense of tightness in the throat. A relationship with Meige's syndrome has been suggested (Marsden & Sheehy, 1982). Less common is the soft, low-volume, 'breathless' speech of laryngeal abductor spasm. Although uncommon, the author has seen laryngeal abductor spasm in the context of neuroleptic-treated chronic schizophrenia, where the 'silly' speech was attributed to mental state.

What may be more common in neuroleptic-treated patients is a tensing of oropharyngeal muscles on talking, resulting in thick speech with slurred, indistinct diction, but little or no disturbance of pitch or inflection. Parkinsonism may contribute to this disorder and it is often difficult to exclude the intruding effects of other established oral dyskinesias, but it is likely that pharyngeal dystonia can present thus more often than is acknowledged.

Segmental dystonias

Many combinations of contiguous involvement can be encountered. Only Meige's syndrome will be mentioned here.

Meige's syndrome

For such a comparatively rare disorder, the syndrome described by Meige in 1910 as 'spasme faciale mediane' (Meige, 1910; Tolosa & Klawans, 1979) is known by a confusing array of names. Perhaps the

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most satisfactory is the descriptive one: 'blepharospasm - oromandibular dystonia syndrome', although the eponym (or one of the several eponyms) is now often applied when only one of the constituent features is present (blepharospasm or oromandibular dystonia). In contrast to the virtual neglect of Meige's paper in the past, the last 15 years have seen a burgeoning of interest in the condition (Paulson, 1972; Marsden, 1976*b*; Tolosa, 1981; Stahl *et al*, 1982; Jankovic & Ford, 1983; Tolosa & Marti, 1988). With increasing recognition it has been suggested that this syndrome may be as common as spasmodic torticollis (Lees, 1985).

Peak onset is in the sixth decade and there is a female excess (Marsden, 1976*b*; Jankovic, 1988*b*). The blepharospasm component is no different clinically from that described above. In Marsden's series of 39 cases of what he referred to as Brueghel's syndrome, a third had blepharospasm alone. Thus this syndrome increasingly appears to be incorporating cases of so-called 'essential' blepharospasm, though as was noted, when examined carefully and repeatedly, the great majority of patients with blepharospasm will eventually show signs of other craniocervical dystonia.

The oromandibular component comprises lip pursing, 'bridling' retraction of the corners of the mouth, forced mouth opening either in the midline or with lateral jaw displacement, tongue protrusion, and spasms of the floor of the mouth. The platysma may also be involved. Spasms are of varying duration and can contort the whole face into a disfiguring expression of revulsion and can be sufficiently violent to dislocate the jaw (Marsden, 1986). Extension to other areas may occur, resulting, for example, in grunts and snorts similar to those found in Gilles de la Tourette's syndrome and arising from involvement of respiratory musculature. Involuntary levator inhibition, torticollis, spasmodic dysphonia from laryngeal adductor spasm and postural tremor are all regularly encountered (Marsden & Sheehy, 1982; Tolosa & Marti, 1988; Jankovic, 1988*b*). The same 'triggers' noted for blepharospasm may precipitate facial contortions which in turn may be alleviated by 'sensory tricks'.

Blepharospasm–oromandibular dystonia syndrome usually has no identifiable cause (primary Meige's syndrome). Increasingly however these features are being noted as secondary phenomena, especially to drug ingestion. The term 'secondary Meige's syndrome' has been applied in this context but the eponym, if used at all, is best reserved for the idiopathic variety (Jankovic, 1988*b*; Burke & Kang, 1988). Neuroleptic drugs are the most commonly implicated pharmacological agents (Weiner *et al*, 1981; Burke *et al*, 1982) but others include L-dopa in the treatment of Parkinson's disease (Weiner & Nausieda, 1982), antihistamine decongestants (Powers, 1982), and amphetamines (Jankovic, 1981). As noted above however (cf. Classification) a range of neurological disorders must also be considered.

Generalised dystonia

Generalised dystonia may be symptomatic or idiopathic. Many, although not all, cases of idiopathic generalised dystonia are familial, with an autosomal dominant pattern of transmission demonstrated in many families (Zeman & Dyken, 1968). Furthermore, in Ashkenazi Jews, who appear to have a higher prevalence of this disorder than other ethnic groups (Fahn *et al*, 1987), an autosomal recessive pattern has been postulated, although this remains somewhat controversial. An X-linked pattern has also been found but only in the Philippines (Lee *et al*, 1976). The dystonia gene may be more widely distributed than has thus far been appreciated, especially if all dystonias are considered as a unified nosological entity.

Idiopathic generalised dystonia is an uncommon disorder, usually beginning in childhood or adolescence and progressing to severe disability. It is unlikely to be encountered in a primarily psychiatric setting.

Drug-related (symptomatic) dystonias

'Tardive' dystonia

The term 'dystonia tarda' (i.e. dystonia associated with prolonged exposure to antidopaminergic drugs) was first used by Keegan & Rajput (1973). These authors were drawing attention to persistent dystonic postures, which, although noted in some previous studies of drug-related involuntary movement disorders, had largely been overlooked. It is only in recent years that 'tardive dystonia' has been given much consideration (Burke *et al*, 1982). This is in striking contrast to the other varieties of involuntary movement associated with long-term neuroleptic exposure.

The general impression is that this disorder is rare (Gerlach, 1979; J. Kane quoted in Burke & Kang, 1988). In one of our studies, using the 43-item Rockland or Simpson Dyskinesia Rating Scale, which allows for rating of at least some dystonic forms, we found that this type of abnormality was recorded infrequently in a sample of 411 chronic schizophrenics (Owens, 1985). Ankle rotation was the most frequently noted abnormality (5.4%), while

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retrocollis, torticollis, and axial dystonia were noted only in 0.7%, 1.5%, and 2.7% respectively. Since 47 patients in this sample had no history of neuroleptic exposure, some of the abnormality included in these figures may represent spontaneous idiopathic disorder. By contrast, the drug-related component may have been under-represented by virtue of the group being relatively elderly (see below). Nonetheless, figures of this order are in broad general agreement with those of Yassa *et al* (1986), who found an overall prevalence of 2% in a similar population using the same rating technique. Such figures, and assumptions about the infrequency of the disorder, must however be viewed critically. Most established dyskinesia rating scales are designed to rate overwhelmingly hyperkinetic-type disorder, not static postural distortions. If the history of other forms of movement disorder in psychotic patients is to be repeated, it is very likely that increasing awareness and skill in recognition, especially of the subtler dystonic manifestations, will lead to greater prevalence rates being reported.

Tardive dystonia does not differ from what has already been described, and the same terminology is appropriate. Axial dystonia imparting a fixed rotational lateral tilt to the trunk, when unilateral, has been referred to as the 'Pisa syndrome' (Ekbom *et al*, 1972; Gerlach, 1979) because of the patient's characteristic truncal lean. The author has also seen bilateral involvement with resultant hyperextension of the spine, accentuated lumbar lordosis, compensatory increase in length of step, and a marked subjective feeling of instability, especially where gait could not be altered, for example when descending stairs. The 'Pisa syndrome ' is not however a separate entity, and the name is best dropped.

The majority of tardive dystonias (perhaps up to two-thirds of cases) begin in the muscles of the face and neck, although most patients subsequently develop disorders elsewhere (Burke & Kang, 1988). Isolated focal craniocervical dystonias appear to be uncommon. At point of maximum severity a segmental pattern of distribution is usual (Kang *et al*, 1986). Retrocollis was a particularly characteristic part of the clinical picture in Kang *et al*'s patients, being present in approximately 40% of cases.

Onset tends to be gradual after months or more commonly years of treatment with dopamine antagonists, although atypical onset after only a few weeks or even days of exposure has been described (Burke *et al*, 1982). As in other contexts, 'tricks' may help and activity exacerbate the disorder. Progression does not appear to be inexorable in that most patients seem to reach a plateau of severity (Burke & Kang, 1988), although outcome in terms of severity, distribution, functional disability or reversibility cannot be predicted. Functional disability is likely to be greater with dystonic than with choreiform disorder.

An important factor in the clinical expression of tardive dystonia is age. Prevalence appears to increase linearly with age (Kang *et al*, 1986). However, it also seems that patients with tardive dystonia alone are younger than those with other manifestations of neuroleptic-related hyperkinesis but no dystonia (Gimenez-Roldan *et al*, 1985; Yassa *et al*, 1986). Hence the cumulative distribution curve for tardive dystonia with age appears 'shifted' to the left compared with that for 'classic' tardive dyskinesia. Furthermore, distribution is to some extent related to age of onset. As with idiopathic dystonia, the younger the subject at onset the greater the likelihood of generalisation (Burke *et al*, 1982; Kang *et al*, 1986). In common with other neuroleptic-related movement disorders, however, there is no proven association with type of neuroleptic, dosage, or duration of exposure. Also there is no known difference in prevalence between the sexes.

Other manifestations of neuroleptic-related movement disorder may be present, such as choreiform, tic-like and myoclonic dyskinesias, as well as tremor. In about half of Kang *et al*'s sample, however, dystonia presented in the absence of a history of orolinguomasticatory dyskinesia.

Neuroleptic-related dystonia remains a relatively 'new' entity. Most of the studies have so far come from neurological centres which are likely to experience differential referral predicated largely on the referring agent's curiosity or the patients' functional disability. Studies with their emphasis on 'bizarre' or disabling kinetic disorder are unlikely to provide a balanced picture of the extent of the problem. It is very probable that a large 'pool' of static dystonic postural abnormalities is lurking within the long-term psychotic population, and psychiatrists must now add this to the differential list of motor disorders, long known but oft forgotten, to be found in such patients (Marsden *et al*, 1975; Owens *et al*, 1982; Rogers & Hymas, 1988).

Other drug-related dystonias

Dystonia as both an early and late manifestation of untreated Parkinson's disease has been known for about a century (Gowers, 1888; Purves Stewart, 1898). It is also however a well described complication of L-dopa therapy of this condition, its emergence apparently relating to the periodicity of the clinical effects of the drug (Poewe *et al*, 1988).

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'Off-period' dystonia is commonest, affecting up to a third of patients treated with L-dopa for longer than three years (Nausieda *et al*, 1980; Curtis *et al*, 1984). It occurs when the drug's effectiveness is wearing off or at its lowest. It is especially likely to occur in the morning before first dose and has a predilection for the legs (Poewe *et al*, 1988). 'Biphasic' dystonia occurs at onset and waning of drug efficacy and tends to have a unilateral distribution, while 'peak-dose' occurs at the height of anti-Parkinsonian activity and most often involves craniocervical areas (Poewe *et al*, 1988). These phenomena do not appear to relate to the duration of treatment but seem to affect those with early-onset Parkinsonism (under 50 years). Management that does not involve some compromise of anti-Parkinsonian efficacy is difficult.

Dystonia occurring with diphenylhydantoin treatment is confined to patients with plasma levels in the toxic range, occurs with other dyskinesias, and is readily reversible with dose reduction (Chadwick *et al*, 1976). Rarely, dystonia has been attributed to carbamazepine administration, although only as a transient or readily reversible phenomenon in severely brain-damaged children (Crosley & Swender, 1979) or young adults (Jacome, 1979). Movements of a more jerky choreoathetoid type have been noted with dextroamphetamine administration in hyperactive children (Mattson & Calverley, 1968).

Psychogenic dystonia

As will be evident from the above, neurologists have in recent years reasserted their traditional suzerainty over the dystonias, largely by unifying a diverse range of clinical disorders into a classificatory system that acknowledges a spectrum of illness. This reassertion is forceful, but not dogmatic, and leaves open the question of whether dystonia can be psychogenic.

Cases of factitious or willed postural disturbance, admitted by the patient, producing appearances similar to those of dystonia, have been described (Lesser & Fahn, 1978; Batshaw *et al*, 1985). If strictly defined as an *involuntary* disorder, however, such cases cannot be considered as authentic dystonia. Rather, they are consciously simulated. The term psychogenic *dystonia* must be reserved for examples of conversion hysteria.

Conversion hysteria causes many psychiatrists great angst. As a concept, it is one of the cornerstones of psychiatry. As a diagnosis it turns out to be wrong with disconcerting regularity. The balance of opinion is that psychogenic dystonia as a manifestation of conversion is very rare. In the largest published series of patients with dystonia, comprising 932 cases from the Dystonia Clinical Research Center in New York, 24 (or 2.6%) were diagnosed as psychogenic (Fahn *et al*, 1987). It is unclear however if this included simulated dystonia. Hence the true figure for 'authentic' conversion reactions may be even lower.

Despite this, surveys suggest that 40% of patients with dystonia are given a diagnosis of conversion hysteria at some point (Fahn *et al*, 1983), although not always by psychiatrists. This need not necessarily be interpreted as meaning that psychiatrists 'get it wrong' 40% of the time, but may be seen as reflecting a different orientation to a complex clinical phenomenon in which psychogenic factors are clearly important in expression, control, and adaptation, if less so in actual cause. The alternative explanation is, of course, that in such cases psychiatrists 'get it wrong' 40% of the time!

The diagnosis of conversion is as much related to characteristics of the diagnostician as it is to the features being diagnosed, and the literature offers little guidance to help the analytically overcommitted offset their backgrounds. Fluctuating neurological signs, such as shifting or non-dermatomal sensory disturbance, inappropriate weakness ('giving way'), pain on light touch or possibly evidence of factitious abnormality elsewhere have been cited (Fahn *et al*, 1983) but are themselves in large part subjective in their evaluation. Perhaps the best parameters are the lack of a normal progression of symptoms with, for example, no phase of 'action' dystonia, and a presentation dominated by static postural disturbance with a relative absence of typical hyperkinesis.

Even here, however, pitfalls arise. The author has seen one patient with sudden evolution of severe torticollis over a few days following a break-in and attempted rape at gun-point in her New York apartment. She felt 'cured' by behavioural and relaxation techniques and was psychologically exceedingly well adjusted after treatment, but clearly showed continuing minimal head rotation with unequivocal scoliosis when examined four months later. She was quite unaware of this. These postural abnormalities were also evident in family photographs two years afterwards.

In the author's opinion, psychiatrists should be highly circumspect in attributing any dystonic abnormality to a purely psychogenic causation.

Diagnosis

Diagnosis of dystonia is based almost entirely on the clinical appearance. Family history is usually unhelpful, except in the rare genetic forms, which are unlikely to present psychiatrically. Nonetheless,

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psychiatrists must bear in mind the range of disorders that can present with dystonia even if precise diagnosis and management of these is best undertaken in a neurological centre. The most important factor for psychiatrists to evaluate is drug history, especially of antipsychotics.

It is essential that psychiatrists remain aware of the unique characteristics of this type of involuntary movement in order to guard against overzealous and often misguided application of analytical theory.

Pathology/pathophysiology

No consistent anatomical lesion has been identified in dystonia. Studying cases of symptomatic hemidystonia, Marsden *et al* (1985) suggest that the underlying problem emanates from a disruption of input from basal ganglia (thalamus, caudate and putamen) to pre-motor and ultimately motor cortex: dystonia is therefore seen as the result of release of pre-motor cortex from thalamic control.

At the neurochemical level a number of theories have been proposed, usually relating to hypothesised disruption of central dopaminergic/cholinergic balance, but no consistent data have as yet emerged to allow construction of a coherent hypothesis. There is evidence that acute dystonic reactions may result from a complex interplay of factors involving pre-synaptic dopaminergic mechanisms (autoreceptors) as well as post-synaptic receptors (Marsden & Jenner, 1980).

Treatment

The treatment of dystonia is unsatisfactory. Evaluation of any treatment programme must take account of the bizarre characteristics of the disorder, the fluctuating intensity of symptoms, the occasional tendency to spontaneous remission and the non-specific benefits of medication and authoritative professional involvement. Evaluation must also involve the use of standardised techniques for staging the disorder, reliably applied. For these reasons such an uncommon disorder is best investigated and managed in a specialist centre where adequate numbers of patients can be studied against suitable controls. Further single case studies will extend our knowledge but little.

Treatment of idiopathic dystonias

Drug treatments

Representatives of virtually every class of centrally acting drug have been tried. Sheehy & Marsden (1982), for example, note 23 preparations tried in their cases of writers' cramp alone (for a comprehensive list of preparations used in different forms of dystonia see Marsden (1986)). Response to medication is unpredictable, that which benefits one case exacerbating or being ineffective in another. This pharmacological variability may have implications for future classificatory systems.

The most encouraging therapeutic development in recent years has been the efficacy of high-dose anticholinergics in both open (Fahn, 1983; Marsden *et al*, 1984) and double-blind (Burke *et al*, 1986) trials. Benzhexol (trihexyphenidyl) introduced in low/average doses of 2–6 mg daily has been most extensively tested. The dose is increased by the order of 2 mg per week to tolerance or significant improvement. Using this slow incremental regime very high doses have been tolerated (Marsden *et al* (1984) report a maximum of 130 mg daily), though with great individual variability. Average doses range from 20 to 40 mg per day. In general, 50% of children and 40% of adults with idiopathic dystonia obtain at least moderate benefit from high-dose anticholinergics (Fahn & Marsden, 1987). The better response in children may reflect the lower prevalence of central side-effects in them than in adults, and hence their greater tolerance to such regimes (Fahn, 1983; Fahn & Marsden, 1987). The theoretical risk of problems with memory and learning, especially in children, has not yet been reported. It is interesting to note the relative infrequency of anticholinergic psychoses with this slow method of dose increment. The only predictor of response identified so far is early initiation of treatment, with a better response claimed for those started on treatment within five years of onset of dystonia (Fahn & Marsden, 1987).

How anticholinergics work in this situation, why high dosage has to be employed, and why it can take months for improvement to become evident are all unknown, but this at least offers some therapeutic prospects for sufferers of an otherwise disabling and distressing disorder.

Psychological/psychiatric treatment

One of the more important therapeutic influences on dystonia is the physician–patient interaction. Patients are often frightened and confused by this uncontrollable disability which behaves so strangely, and may themselves believe, or may have been told by doctors, that it represents a psychological disorder. Initial medical reaction is important, and should referral be to a psychiatrist it is essential that, before plunging into the recesses of a supposedly disordered subconscious, the almost exclusively

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medical nature of the problem is openly acknowledged. Patients are invariably more stoical in dealing with the disabilities of an illness whose mysterious origins and empirical treatments are admitted than they are at coping with the tyranny of an imposed psychopathology.

This must not be taken as meaning that psychological/psychiatric interventions have nothing to offer. This is certainly not the case, and in this regard neurologists, by their own admission, may have swung the pendulum too far the other way, as hinted by Fahn *et al* (1987).

A range of psychological treatments have had, and still have, their advocates. The problem is that most reports of efficacy in this field concern single case studies or small patient groups. No controlled trials or long-term follow-up studies of adequate samples have been undertaken. Nonetheless, substantial improvements have been reported with a range of mainly behavioural techniques (Lees, 1985; Faulstich *et al*, 1985; Fahn & Marsden, 1987). The details of these are beyond the scope of this article, but include relaxation (in its many forms), negative practice, and biofeedback. It is likely that the beneficial effects of such programmes lie less in the theoretical constructs behind them than in their non-specific influences on mental state, muscle control, composure, and overall ability to cope. Whatever their mode of action and efficacy – and usually in the context of an acknowledged medical disorder – they are certainly worth trying in skilled hands, when other treatments are of such limited value.

It is perhaps axiomatic to state that any coexistent psychiatric illness should be conventionally managed. Supportive psychotherapy is also a helpful adjunct (Fahn & Marsden, 1987) by providing education, advice, and understanding where all of these may be lacking elsewhere. Prolonged or interpretative psychotherapies have no place in management in the absence of clear psychogenesis. Mostly these are unhelpful, although in some cases where the medical nature of the disorder has not been recognised and treatment has consisted of a stubborn insistence on the development of dynamic insight, they have been clearly detrimental (Eldridge *et al*, 1969; Fahn & Marsden, 1987).

Surgical treatment

Detailed consideration of this highly specialised area is beyond the scope of this article. Surgical procedures range from myectomy, through peripheral nerve and spinal root section, to thalamotomy. Results are variable (Fahn & Marsden, 1987).

An interesting recent innovation, especially in the treatment of blepharospasm, is local (supraorbital) injection of botulinum A toxin (Jankovic, 1988*c*). This is the most potent biological toxin known and was originally introduced in 1980 to treat strabismus. It acts by rapidly and irreversibly binding to presynaptic cholinergic membranes, from where it is internalised, thereby disrupting calcium influx (Jankovic, 1988*c*). Good results have been achieved with few local side-effects, although treatment may have to be repeated.

Treatment of 'tardive' dystonia

Tardive dystonia presents a singular management problem. This relates to the greater functional disability than that associated with other drug-related dyskinesias, the likelihood of sufferers being younger, potential irreversibility and the requirements for long-term medication to maintain psychiatric well-being.

The crucial first-step is a re-evaluation of the need for on-going neuroleptic treatment. A feature of American work is the relatively high proportion of patients with tardive dystonia who had been treated with neuroleptic drugs for non-specific and non-psychotic psychiatric states (Burke *et al*, 1982): 22% of Burke & Kang's (1988) patients had received these drugs for anxiety, multiple somatic complaints, nausea, etc. The situation would (hopefully) be rather different in the UK. Neuroleptics are indicated for non-psychotic states in extreme circumstances only and should be discontinued if dystonia ensues.

Most patients, however, will be schizophrenic. Stable, functioning patients with a history of only one episode may not benefit from prophylaxis (Crow *et al*, 1986) and may therefore survive discontinuation of antipsychotics. Since it is not possible to identify relapsers in this group (MacMillan *et al*, 1986) such patients who develop dystonia are entitled to a trial drug-free period to assess both the response of their neurological signs and their relapse potential. It is important to note that Burke & Kang (1988) have reported that the only patients in their series (with mixed diagnoses) whose dystonia resolved completely came from the ranks of those who came off, and could remain off, antidopaminergic medication.

For those with an established pattern of relapse on neuroleptic withdrawal discontinuation is hardly an option, even with such a problem as dystonia. It is then worthwhile to re-evaluate dosage, and reduce to the minimum to prevent relapse. As noted, there is no evidence linking dystonia to antipsychotic drug dosage, but such a step would seem clinically reasonable if not scientifically logical. It is certainly good practice. Because of the delay of any possible beneficial effects from such a move 'filtering through'

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with depot preparations, the author prefers if possible to switch to an oral preparation in this situation, although this is a personal rather than a proven manoeuvre.

If dystonia persists, specific interventions may be necessary. High-dose anticholinergic as described above may produce improvement in up to half of cases (Kang *et al*, 1986; Burke & Kang, 1988). Exacerbation of coexisting 'classic' (choreoathetoid) tardive dyskinesia with anticholinergic treatment seems more a theoretical than a practical risk, as confirmed in Kang *et al*'s (1986) series, where only one of ten patients showed exacerbation of complex oral dyskinesias.

Alternatively addition of a pre-synaptic catecholamine-depleting agent such as reserpine or tetrabenazine may be helpful (Burke & Kang, 1988) but side-effects such as depression or disabling Parkinsonism may limit their usefulness. Nonetheless, Kang *et al* (1986) found these to be the most useful agents in their dystonic group, although two-thirds of this sample did not have a clear psychotic diagnosis. Depleting agents may be a less practical option in a purely schizophrenic group.

Finally, in 'classic' tardive dyskinesia, increasing the dose of antidopaminergic medication may suppress the movement disorder (American Psychiatric Association, 1979; Jeste & Wyatt, 1982), and where all else fails, this may be considered with drug-related dystonia. No published work to date has addressed this. The theoretical risk of such an approach is that of subsequent 'breakthrough' of abnormal movements at a later date, possibly with greater severity. This is based on assumptions drawn from the post-synaptic dopamine receptor supersensitivity theory of the pathophysiology of tardive dyskinesia. This is of course only a theory which accommodates some, but not all, of the clinical observations concerning drug-related movements, and it is one that has been challenged (Jeste & Wyatt, 1981). To date there is no clinical evidence that this 'breakthrough' actually occurs. Nonetheless, such a strategy is probably best reserved for cases of severe disability or generalised dystonia when all else has failed. When this degree of disorder is associated with evidence of muscle damage (with elevated levels of creatinine phosphokinase) one is dealing with a potentially life-threatening situation (Lazarus & Toglia, 1985; Burke & Kang, 1988) and extreme measures are justifiable.

Conclusion

Dystonia has recently undergone a substantial revision in the neurological literature. The purpose of this article has been to highlight this to a psychiatric audience who, by tradition and inclination, may tend to see this disorder from a different perspective. Knowledge remains scant and it would be wrong to assume that neurologists have a monopoly of insights into these perplexing states. Nonetheless, as Lees (1985) has pointed out, although the evidence linking the disparate presentations of dystonia to each other and ultimately to disorder of the basal ganglia is circumstantial, it is nonetheless persuasive. Psychiatrists would do well to be familiar with it.

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D. G. Cunningham Owens, MD, FRCP, FRCPsych, *Consultant Psychiatrist, Scientific Staff, Medical Research Council, Northwick Park Hospital and Clinical Research Centre, Watford Road, Harrow, Middlesex HA1 3UJ*

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