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Case Conference

Abnormal Involuntary Movements

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The differential diagnosis of an athetoid arm movement in a 54-year-old lady with a psychiatric history of affective disorder was discussed. Further investigations after the conference yielded a firm diagnosis that unified both physical and mental symptoms.

Objective

This is a report of a special case conference at the Royal Liverpool Hospital on 28 November 1988. The conference was asked to examine a patient's physical symptoms in the light of her psychiatric diagnosis.

Presentation of case

Miss D, a 54-year-old, Roman Catholic, unemployed spinster was admitted to the Royal Liverpool Hospital in October 1988 following a consultant domiciliary visit. Her presenting complaints were of abnormal involuntary movements of her right hand and arm, racing thoughts, 'high' mood, and poor sleep. Two weeks earlier she had been complaining that she was depressed because of continuing involuntary movements of her right arm. At that time she stopped her maintenance medication of chlorpromazine, which she had been taking for over five years for bipolar affective disorder. She had previously discontinued her lithium prophylaxis in July 1988 because of worsening psoriasis, and because she felt that her thinning hair was attributable to lithium.

After she stopped the chlorpromazine she rapidly became elated. She became overactive and garrulous. Her sleep had reduced to four hours per night. Her appetite was poor and she was losing weight.

The involuntary movements of her right hand and arm had begun two years earlier. At first she noticed only small movements of the wrist and fingers, but these became gradually worse so that they eventually involved the whole arm and hand in athetoid movements. These increased with anxiety and improved if she was distracted or relaxed. On some days the movements would be worse than others. The athetosis could sometimes wake her at night. When sitting in a chair she would hold a table with her right hand in order to try to suppress the movement. Just before her admission she felt the athetosis was worse than ever before.

Psychiatric history

In 1977, when she was 42, Miss D was admitted to Rainhill Hospital for six months. The admission followed a series of arguments with her sister and half-sister. She had been rivalling her sister for the attention of her two teenage nephews, and had been interfering in the running of her sister's house, where she lived. After Miss D had searched through her sister's private letters to try to establish that the boys' mother was "unfit to care for them", a solicitor was consulted. In this atmosphere of increasing hostility, Miss D's behaviour became disturbed. She assaulted her half-sister and was admitted from casualty after presenting a mixed affective picture with agitation, low mood, some pressure of speech and flight of ideas, and ideas of guilt and persecution. She complained of "whispering noises" against her in her home.

On the ward she behaved in a variety of abnormal ways, usually at times when she knew she was being observed. She pulled other patients' hair, kicked doors, threw the fire extinguisher, and lay under her bed at night, saying the floor was "softer" than the mattress. She would stand silent and still for minutes holding a hatpin in one hand and a ball of wool in the other. She sang loudly in the evenings. When asked why, she replied, "because it's cheaper after six", a reference to a telephone commercial. On one occasion, when asked how many legs a horse had, she answered by getting down on all fours and whinnying.

For reasons that are not clear in the case notes, a diagnosis of hysterical conversion syndrome was made at that time and she was treated with multiple abreactions using diazepam. Miss D became so suggestible that she eventually would enter a deep trance if only 1 ml of distilled water was injected. Each abreaction focused on her feelings of loneliness and rejection by her sisters. It became clear that she had not mourned her father, who had died in 1972, and she began then to grieve for him. She was discharged well after six months and followed up for two years in the out-patients department on no medication. She remained well.

In 1983, when aged 48, she was admitted to the Royal Liverpool Hospital. Before her admission she had been ostracised by her colleagues at a city-centre department store for refusing to join a union. This, together with uncertainties over the store's future, had weighed heavily upon her. She presented with a history of poor sleep, low mood, tiredness, and aggression towards her half-sister. An initial diagnosis of reactive depression was modified when after one week she became elated, with pressure of speech and flight of ideas. She began overspending, and was interfering and restless. She sang when she had blood taken. She was diagnosed as having bipolar affective disorder and treated with chlorpromazine and lithium. She was discharged two months later on chlorpromazine (25 mg b.d. and 125 mg *nocte*) and lithium carbonate (800 mg *nocte*). There were no features of akathisia or dystonia noted then. She was maintained on varying combinations of these two drugs until 1988. In the follow-up clinic over these five years she was noted to become elated only twice, and these episodes coincided with a fall in her serum lithium.

Medical history

Apart from a groin abscess as a child and psoriasis from young adulthood her physical health was good until 1985, when she had a right mastectomy for breast cancer. She was given radiotherapy and tamoxifen. On admission her only regular medication was tamoxifen (20 mg b.d.).

Family history

Miss D's Roman Catholic mother died aged 42 of ovarian carcinoma, when Miss D was eight. At the time of her death her mother was pregnant with twins.

Miss D's Protestant father brought up the family's three children. He became a "father, mother and brother" to Miss D. He was a self-employed plumber. Miss D was conscious that he would have preferred her to have been a boy to act as his 'right hand man' in his business. He died of pneumonia in 1972 aged 85.

She had two siblings: an elder half-sister, N, aged 61, from her mother's previous marriage; and C, a sister aged 52, who had two illegitimate children. All the children were brought up as Catholics. The three sisters were very close, and lived together until 1965 when C became pregnant and had to leave the family home.

There was no psychiatric illness or similar movement disorders in the family.

Personal history

Miss D had a normal birth and was brought up in Liverpool. She had a normal, happy infancy, living with her father, mother, and two sisters. She had no neurotic traits. After her mother died, her father became her main emotional support. Miss D went to school during the war at two inner-city Catholic schools. Unlike many of her generation in Liverpool she was not evacuated. She left school at 15 with no qualifications and became a shop assistant for two years. At the age of 17 she joined the WRAF, "to prove to my father that I was as good as a boy. There had been generations of our family in the Army for 200 years." After this she spent 20 years as a stockroom supervisor for a chain store.

After 1970 she began to help her sister, C, look after her two illegitimate sons. Her father died in 1972. In 1977 she became 'overinvolved' with her two nephews and suffered her first psychiatric illness, but returned to work some six months later. She stopped work in 1983 when the store closed, and has since remained unemployed.

At menarche she was 12. Her half-sister N had already told her the 'facts of life'. From the age of 13 her sexual fantasies were predominantly heterosexual. Her main mode of sexual gratification was, and is, masturbation. Miss D's strict Catholic upbringing is reflected in the fact that at 53 she is still a virgin. However, she had been engaged three times, at 17, 19, and 38. Her first engagement lasted only a few months because she refused to have full sexual intercourse. Her second engagement, when she was 19, lasted ten years. Again, the relationship did not progress beyond foreplay, and the engagement ended when he suddenly married someone else. When she was 38 she became engaged for the last time, but this ended when her Protestant fiancé became a vicar. She nearly became engaged for a fourth time when she was 39. She wears a dress ring on the ring finger of her left hand, which she describes as a present to herself when she was 40. It looks just like an engagement ring. She has no boyfriend now.

Social circumstances

Miss D lives with her elder half-sister, N, in their jointly owned house, which is the family home in which she was born. The household is financially stable. Miss D smokes 200 cigarettes a week, but drinks no alcohol.

Pre-morbid personality

Miss D is normally said to be jolly, chatty, and 'bubbly'. She maintains an optimistic outlook on life, and rationalises her misfortunes away. She admits to being sensitive to criticism and sometimes lonely. In her work she was a conscientious perfectionist, and keeps this attitude in her housework, which is carried out according to a rigid routine. She still attends Mass regularly. She has few hobbies, and regards herself as the mainstay of the family. There is no evidence of pre-morbid cyclothymia.

Mental state examination

Miss D is a 54-year-old caucasian woman, who looks somewhat older than her years. Her hair is neglected and thinning but otherwise she is clean and tidy. Both hands have nicotine-stained fingers and cherry-red finger nails which she had painted herself to conceal psoriasis.

During the interview Miss D was co-operative and maintained excellent rapport. She was slightly agitated. Her right hand and arm were involved in gross athetoid movements.

As the patient relaxed through the interview the movements became less marked.

Objectively Miss D was cheerful to the verge of elation. She became tearful suddenly in the interview when discussing her late father. Subjectively she said her mood was normal for her. She was garrulous and difficult to keep to the point. Her speech was relevant but tended to circumstantiality.

The form of her thoughts was normal, but the content was dominated by her pre-occupation with the athetosis of her right arm. There was a slight grandiose flavour to her ideation – she believed that indirectly her admission for assessment would benefit "a great many people", but there were no overt psychotic features. She was cognitively intact and understood the purpose of her admission. She felt her athetosis was due to the chlorpromazine.

Physical examination and investigations

Miss D was a thin, elderly lady with a focal athetosis. Her hair was noticeably sparse. She had a conjunctivitis. There was a right mastectomy scar, but no evidence of recurrence at the site or lymphadenopathy.

There was no evidence of orofacial dyskinesia. A neurological examination was normal except for the athetosis, associated with weakness and increased tone. There was some incoordination with dysdiadochocinesia of the right hand.

A full blood count, urea and electrolytes, liver function tests, thyroid function tests, syphilis screen, chest and cervical spine X-ray, computerised tomography (CT), and EEG were all normal. There were autoantibodies to cell nuclei. Her serum copper level was mildly raised.

Course on admission

Miss D was re-started on her lithium, but not her chlorpromazine. Tetrabenazine was added to the lithium and tamoxifen. A month after admission, although the athetosis persisted, the patient reported a definite improvement in the movements. Her mental state was then normal and she had returned to her usual enthusiastic and chatty self. A neurologist saw her in hospital and diagnosed the condition as tardive dyskinesia.

Conference discussion

DR POOLE (*consultant psychiatrist and chairman*): This is a complicated case and several questions arise. I find it hard to accept the neurological opinion that this lady's abnormal movements are due to tardive dyskinesia; furthermore, the treatment of her psychiatric state and neurological problem interfere with each other. We must also bear in mind the significant psychological issues, particularly those of grief and loss.

To facilitate the proceedings, a possible differential diagnosis, produced at the time of admission, was presented for discussion. Five causes of the abnormal movement were put forward: tardive dyskinesia, a discrete lesion of the left basal ganglia, a secondary neoplasm, hysterical conversion, or some other systemic condition such as multiple sclerosis, Wilson's disease, or Huntington's chorea.

DR ABOU-SALEH (*senior lecturer in psychiatry*): As regards diagnosis this is undoubtedly a case of focal athetosis of the right arm, and although bipolar affectively ill patients are highly vulnerable to tardive dyskinesia, there is no evidence of tardive dyskinesia itself. The aetiology of this condition may or may not be related to exposure to long-term neuroleptic treatment: it may be a coincidental development, and this is more likely as it is unilateral and circumscribed and it is not associated with tardive dyskinesia. One should also mention, if only to dismiss it, the possibility of a cerebral tumour secondary to breast cancer. The CT scan was negative, but I wonder whether nuclear magnetic resonance with its higher resolution could show a small lesion?

As regards management, tetrabenazine is the most appropriate treatment, but this could complicate the management of the affective disorder: its powerful amine-depleting effects might precipitate a depressive relapse. Lithium appears to have been effective except for aggravating her psoriasis, and the alternative might be carbamezapine.

DR WILLIAMS (*senior house officer*): I notice from interviewing the patient that she has perfectly polished red nails which she has been able to do herself, using both hands in turn. She says she has always been able to control her hands to paint her nails, even when the movement was at its worse.

Tardive dyskinesia can fluctuate in its severity according to the patient's level of arousal: the patient might still have tardive dyskinesia and, when relaxed, be able to control hand movements enough to apply nail polish.

The commonest site for tardive dyskinesia is around the mouth and tongue, and this is absent in this patient. There is support for involvement of the trunk, arms, hands and legs forming a distinct subgroup of tardive dyskinesia (Kidger *et al*, 1980). The elderly are seen as being more vulnerable to developing tardive dyskinesia on neuroleptic medication and usually the form taken is that of a choreiform movement (Task Force on Late Neurological Effects of Antipsychotic Drugs, 1980). Recent work on the pathophysiology of the condition suggests that chronic dopamine blockade of striatal neurons produces altered activity in the striatonigral GABA pathway resulting in irregularities in GABA transmission from the basal ganglia (Thaker *et al*, 1987).

Tardive dyskinesia is however seen as a diagnosis of exclusion. Even when there is a history of prolonged neuroleptic administration, other causes of persistent movements should be ruled out (Munetz & Benjamin, 1988).

DR LOVETT (*lecturer in psychiatry*): I think Miss D's presentation may be due to multiple aetiologies, which need not be mutually exclusive. She may indeed have a bipolar affective diorder, but this does not exclude the possibility that the 'involuntary' movement of her right arm is the result of a hysterical conversion state. There is evidence in her past psychiatric history that she uses dissociative mechanisms to cope with stress. Alternatively the movements could be the result of hysterical elaboration of an organic condition which is yet to be identified.

There are suggestions in her psychosexual history and psychiatric history that hysterical behaviour is not uncommon in this lady, and perhaps hysterical elaboration is in keeping with her personality. Whether she has a full-blown hysterical personality disorder is more difficult to say, but of course a hysterical conversion state can occur in any personality type.

Miss D's hysterical conversion state was diagnosed in 1977, and six years later she was given a different diagnosis. This reminds one of Eliot Slater's work on patients given the diagnosis of hysteria in the 1950s and followed up nine years later (Slater, 1965). He found that of the 99 patients he could follow, only 19 were symptom free. There had been a missed organic diagnosis in a third. Such organic diagnoses as disseminated sclerosis, neoplasms in the central nervous system, vascular disease, epilepsy, dementia, cord compression, and trigeminal neuralgia were overlooked. There were also missed psychiatric diagnoses of obsessional neurosis, schizophrenia, and bipolar affective disorder. Only seven patients at follow-up could be said to have had a true conversion syndrome.

DR HIGGO (*registrar*): Slater's work on hysteria related to work at the National Hospital in Queen's Square

(a neurological unit) and might therefore not be typical. Lewis (1975) showed that the majority of patients with hysteria were completely well on follow-up and thus did not have serious organic disease which had not been diagnosed. Some patients still had symptoms closely related to their original presentation. Reed (1975) showed that a majority of patients had genuine psychiatric disorders, or could be called histrionic personalities.

Overall I would suggest that 'hysteria' frequently represents significant psychiatric morbidity. It is incorrect to suggest that 'hysterics' merely have undiagnosed physical illness. Perhaps we should rather see them in terms of abnormal illness behaviour.

DR POOLE: This lady's history is marked by a pattern of grief and loss. It would appear that she uses mechanisms of manic denial, and the mixed affective picture might be understood in this context. You do not necessarily need to invoke the diagnosis of hysterical conversion to accept that these movements may be resolving many different questions for this lady, and thus may be exacerbated and sustained by the mechanism of secondary gain.

DR UPADHYAYA (*lecturer in psychiatry*): I feel that a lesion of the left basal ganglia is the most likely differential diagnosis for the athetosis. It may be that the CT scan did not pick this up. Further brain imaging such single-photon emission tomograms or magnetic resonance imaging could reveal abnormalities in the basal ganglia. These are sometimes very difficult to find. A brief report by Besson *et al* (1988) showed a lesion only on studies of cerebral perfusion in the striatal area. They suggested that such abnormalities indicate a favourable response to drug treatment for torticollis.

PROF. COPELAND (*head of department of psychiatry*): From what you have told us there does not seem to me to be any real evidence that the patient had an hysterical illness in the past. All her symptoms in 1977 seem to be consistent with a mixed affective state, so I would tend to dismiss that diagnosis.

Is this tardive dyskinesia? She has several risk factors: age, female sex, and antipsychotic medication. Kane (unpublished) claimed that affective disorder in schizophrenic patients carried an increased risk. However, as Dr Abou-Saleh pointed out, the picture is atypical in being unilateral and confined to the arm, although, rarely, this can happen. The danger here is to assume that the neuroleptics have brought about the condition, and to overlook a neurological cause.

One is looking for disorders which cause these symptoms, either alone or in combination with psychiatric symptoms. Some we have mentioned, but there are others which are rare. I do not know of any presentations of Wilson's disease or Huntington's chorea which resemble this patient's illness. You will need to persevere with investigations, but often no cause is found.

Regarding treatment, I do not have much to add. If you conclude that this is tardive dyskinesia, there have been several reports of the value of calcium channel blockers (Ross *et al*, 1987; Leys *et al*, 1988), but I do not have any experience of using them myself. I think that tardive dyskinesia is not the diagnosis.

Conclusion

Several abnormal investigations were returned after the case conference. These included a heat inactivation test and positive DNA autoantibody tests. These tests were reviewed and repeated by a consultant rheumatologist. Taking the clinical history as a whole, the complaint of alopecia, and reduced tear production on examination, he was able to conclude that this patient had systemic lupus erythematosus (SLE). This has important implications. The abnormal arm movements might then be seen in the context of discrete cerebral microinfarcts, undetected by CT. (We await the results of nuclear magnetic resonance imaging.)

The excess incidence of neuropsychiatric illness in patients with SLE is well documented and may reach 60% of SLE sufferers. Such neuropsychiatric manifestations can mimic the clinical pictures of hysteria, schizophrenia, and bipolar affective disorders (Feinglass *et al*, 1976). The diagnosis of SLE with involvement of the central nervous system might well provide a complete explanation for the differing psychiatric presentations in this patient and the abnormal arm movements.

A number of drugs are capable of producing serological and clinical features of SLE. These drug-induced lupus syndromes can be caused by anticonvulsants (phenytoin, ethosuximide), antiarrhythmics (procainamide), antihypertensives (methyldopa), and antibiotics (penicillin, sulphonamides). Chlorpromazine, which this patient received for five years, is also included (Weinstein, 1980). However, on the basis of the repeat tests he had performed, the consultant rheumatologist confirmed the diagnosis of SLE.

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