Severe persistent chorea with phenothiazine therapy: report of a case

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PERSISTENT DYSKINESIA affecting the face, mouth, tongue and jaw (oral dyskinesia) is well known as an insidious and late development of protracted administration of phenothiazines. The syndrome has been reviewed by Crane (1968) and Kline (1968). Some of the patients described also showed dyskinesia affecting the neck, limbs and trunk, manifesting usually as generalized restlessness or minor choreiform movements at the extremities (Hunter, Earl & Janz, 1964; Faurbye *et al.*, 1964; Pryce & Edwards, 1966). More violent choreiform movements may rarely occur (Druckman, Seelinger & Thulin, 1962; Kennedy, 1969). These cases are easily distinguishable from Huntington's chorea because of the presence of prominent and characteristic oral dyskinesia, apart from the lack of family history. However, descriptions of individual cases in the literature have been sketchy and we are not aware of any report where marked choreiform movements of limbs and trunk occur in the presence of only minor and unobtrusive oral movements. It therefore seems worthwhile describing such a case. Because of co-existing dementia this case was clinically indistinguishable from Huntington's Chorea.

Case report

A Chinese man of 37 was diagnosed paranoid schizophrenia at the age of 19 in 1952. He has since been treated with phenothiazines (chlorpromazine, perphenezine, trifluoperazine and trifluopromazine) given singly at different times in moderate doses. He also had a course of deep insulin coma, and in 1955 was leucotomized without postoperative sequalae. In 1964 at the age of 31, he had a tremor affecting the tongue, lips, shoulders and hands. The movements gradually became more severe and choreiform spreading from the face to the neck, limbs and trunk. Artane up to 4 mg t.i.d. did not improve him. In July 1969, both chlorpromazine and Artane were stopped. By this time the movements were violent and substantially impaired his manual dexterity, locomotion and speech. They were most prominent in the proximal parts of the upper limbs and were quick, jerky, complicated, irregular and varied. They were aggravated by mental exertion, diminished by voluntary effort for several moments at a time and disappeared in sleep. Movements involving the face consisted of lip twitching and pouting but these were minor and quite overshadowed by the limb, trunk and neck movements. His affect was blunted, grasp poor and he was retarded and perplexed, the picture being one of organic psychosis. His physical condition was otherwise good. Liver function tests, EEG, caeruloplasmin and urine copper were normal. He had no previous history of encephalitis or other neurological disorder. Family history was negative for chorea or other neurological illness. His condition remained unchanged 10 months after withdrawal of phenothiazines.

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Discussion

Most of the 'irreversible' dyskinesias and involuntary movements after phenothiazine administration reported in the literature were found to be easily reversed with anti-Parkinson agents (although some were made worse) and discontinuing the phenothiazine. The involuntary movements in this patient were unaffected by anti-Parkinson medication and persisted unchanged 10 months after withdrawal of chlorpromazine. Kline (1968) has suggested that 6 months after withdrawal of drugs would be a reasonable cut-off point in assessing 'irreversibility' in the case of oral dyskinesia.

The question of a cause-and-effect relationship between persistent dyskinesias and phenothiazines has not been finally resolved. Such a relationship is suggested by the known effect of phenothiazines on the extrapyramidal system. Also phenothiazines may induce fits in the brain-damaged and increase the frequency of fits in the epileptic, that is, presumably cause or aggravate brain damage. Pryce & Edwards (1966) showed that the development of oral dyskinesia was directly related to the dosage of

phenothiazines. Edwards (1970) showed that persistent oral dyskinesias in elderly women in mental hospitals were associated with a high incidence of brain damage. It is probable therefore that prolonged phenothiazine intake and leucotomy in the present case contributed to the development of chorea and dementia.

Cases like the present one if presenting with chorea for the first time may be mistaken for Huntington's chorea where a history of phenothiazine intake is lacking owing to the demented state of the patient. Phenothiazine-induced dyskinesia should therefore be considered in the differential diagnosis of chorea of unknown origin. The diagnosis of this dyskinesia is also important in view of the finding by Roxburgh (1970) of a good response of the condition to thiopropazate hydrochloride.

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References

CRANE, G.E. (1968) Tardive dyskinesia in patients treated with major neuroleptics. *American Journal of Psychiatry*, **124**, Suppl., 40.

DRUCKMAN, R., SEELINGER, D. & THULIN, B. (1962) Chronic involuntary movements induced by phenothiazines. *Journal of Nervous and Mental Disease*, **135**, 69.

EDWARDS, H. (1970) The significance of brain damage in persistent oral dyskinesia. *British Journal of Psychiatry*, **116**, 271.

FAURBYE, A., RASCH, P.J., PETERSEN, P.B., BRANDBORG, G. & PAKKENBERG, H. (1964) Neurological symptoms in pharmacotherapy of psychoses. *Acta psychiatrica Scandinavica*, **40**, 10.

HUNTER, R., EARL, C.J. & JANZ, D. (1964) A syndrome of abnormal movements and dementia in leucotomized patients treated with phenothiazines. *Journal of Neurology, Neurosurgery and Psychiatry*, **27**, 219.

KENNEDY, P.F. (1969) Chorea and phenothiazines. *British Journal of Psychiatry*, **115**, 103.

KLINE, N. (1968) On the rarity of 'irreversible' oral dyskinesias following phenothiazines. *American Journal of Psychiatry*, **124**, Suppl., 48.

PRYCE, I.J. & EDWARDS, H. (1966) Persistent oral dyskinesia in female mental hospital patients. *British Journal of Psychiatry*, **112**, 983.

ROXBURGH, P.A. (1970) Treatment of persistent phenothiazine-induced oral dyskinesia. *British Journal of Psychiatry*, **116**, 277.