

Treatment of writer's cramp with sodium valproate and baclofen

A case report

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Summary

Treatment of a 27-year-old Black man with writer's cramp with a combination of sodium valproate (Epilem) and baclofen (Lioresal) resulted in dramatic improvement of symptoms and signs. The possible mechanism of action of these drugs is discussed. This combination should be tried in the initial management of this syndrome.

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Writer's cramp is a condition characterized by muscle spasm, inco-ordination and discomfort, variously described as fatigue, weakness, stiffness or pain during an attempt to write.¹ Although the nature of this condition is still debatable, many authors believe that the condition represents a focal form of dystonia.² Indeed some patients with dystonic writer's cramp clearly have dystonia elsewhere, such as torticollis.³ However, when the condition is confined solely to the act of writing (simple writer's cramp) it must be treated as a separate problem.

The treatment of writer's cramp has been unsatisfactory so far. Since drugs with dopamine receptor-blocking properties (e.g. haloperidol, reserpine, tetrabenazine) have been used with limited success in the treatment of the dystonic spasm,³ it is possible that the syndrome results from a striatal dopaminergic predominance.⁴ Recently Brennan *et al.*⁵ reported good clinical response in a severely affected patient with focal dystonia (Meige's disease) treated with a combination of sodium valproate (Epilem) and baclofen (Lioresal). I report on a man with simple writer's cramp, not previously treated with neuroleptic agents, in whom complete and sustained remission of signs and symptoms resulted from 4 weeks of treatment with the gamma-aminobutyric

acid (GABA)-mimetic combination of sodium valproate and baclofen.

Case report

A 27-year-old Black man was referred to the medical outpatient department with 1 year's history of writing difficulties. He reported that while writing his arm and forearm would become tense and painful, his palm would perspire, tremor would occasionally appear, and he tended to lose the tight grip of his pen. His symptoms had progressed over the past few months; his writing became slow and laboured after 1 or 2 hours. Finally he would stop to rest and try again. The cramp was characterized by the overriding pressure of his adducting thumb, followed by the pen tending to roll out of his tight grip.

He appeared to be a serious-minded, intelligent, cautious and precise individual, not unduly depressed. Physical examination revealed no abnormality apart from his writing difficulty. In his attempts to write the patient tended to over-flex and adduct his thumb, and extend his second and third fingers while tightly gripping his pen. His writing was jerky, his hand perspired excessively, and his wrist would flex and elevate above the writing surface. In addition, his arm and forearm appeared tense. No other neurological deficits were detected. An extensive investigation including routine serum biochemical estimations, a full blood count, thyroid and liver function tests, serum ceruloplasmin, vitamin B₁₂ and folate estimations, skull radiographs, electro-encephalography and computed tomography scans revealed no specific cause for the dystonia. A diagnosis of writer's cramp was made.

Treatment with sodium valproate and baclofen was started, initially at 600 mg and 30 mg respectively in 3 divided doses daily. Over a period of a week the daily doses were increased to 1 200 mg and 60 mg respectively. Two weeks after treatment the patient's writing gradually improved, and noticeable remission of signs and symptoms was noted after 4 weeks. The condition remained in remission over a 3-month period and no side-effects or toxic effects of the drugs were noted.

Discussion

The pathophysiology of writer's cramp is unknown. Since the syndrome represents a focal form of dystonia, it might result from a striatal dopaminergic predominance, secondary to disin-

hibition of dopamine striatal neurons. The alleviation of the spasm, although usually transient, by drugs that block dopamine receptors tends to support this notion.⁴

Sodium valproate is thought to act by increasing the synaptic concentration of GABA in the brain; baclofen has recently been shown to be a selective agonist for GABA_B receptors which are present on central nerve terminals and through which the release of several transmitters, including dopamine, seems to be inhibited.⁶ A combination of the two drugs could therefore be expected to act synergistically, reducing activity in the nigrostriatal dopaminergic pathway and inhibiting release of dopamine from dopaminergic terminals in the striatum. It is therefore suggested that such a combination of actions might represent a more physiological means of reducing dopaminergic predominance in the striatum than direct blockade of dopamine receptors.

In view of the well-known complications of long-term administration of neuroleptic agents and the relative inefficiency of these drugs in relieving writer's cramp, it is believed that the combination of sodium valproate and baclofen should be tried in the initial management of this condition.

Addendum

For the past 6 months I have been treating a 42-year-old accountant for dystonic writer's cramp. Initially the patient was able to write 2-3 sentences and was forced to stop writing owing to a cramp-like sensation at the wrist and proximal arm. After 3 months' treatment with sodium valproate and baclofen he was able to write up to 2 pages without experiencing any discomfort. These 2 reported cases confirm that writer's cramp is probably a form of focal dystonia in which a deficiency of the central neurotransmitter GABA plays an important role.

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