Tardive Lingual Dystonia Treated with Clozapine

Tardive dystonia is associated with long-term use of dopamine D_2-like receptor blocking agents, such as typical neuroleptic drugs (1). Tardive dystonia rarely remits spontaneously; clinically, it is indistinguishable from idiopathic torsion dystonia and mainly presents with a focal or segmental distribution (2,3). It has been observed recently that tardive dystonia improves when typical neuroleptics are replaced by clozapine, an antagonist of dopamine D_4 receptors (4,5). The efficacy of clozapine on tardive dystonia is to be weighed against alternative therapies, such as botulinum toxin. However, the latter usually is not injected into lingual muscles because of the possibity of inducing choking. We observed a patient who developed severe tardive lingual dystonia after a chronic treatment with haloperidol and rapidly improved when treated with clozapine.

A 22-year-old male schizophrenic patient who had been treated with haloperidol decanoate (150 mg every other week) for 7 years was first seen by us in 1992. Past medical history and family history were unremarkable for psychiatric or movement disorders. Dystonic movements of the mouth and tongue first occurred 2 years after starting neuroleptic treatment. Anticholinergic drugs (trihexyphenidyl 2 mg three times daily) provided relief for some months; then lingual dystonia progressively worsened. Gradually, lingual dystonia became extremely severe.

Language was often unintelligible and could be ameliorated by the sensory trick of putting a large handkerchief in the mouth. Eating became difficult, and meal time was protracted; in order to stop food from slipping out of his mouth, the patient had to push it with his fingers. Solid food often caused choking, and liquids had to be gulped down.

Attempts to decrease neuroleptic drugs were unsuccessful due to the reappearance of behavioral disturbances. Increase of trihexyphenidyl up to 30 mg/day provided no relief on dystonia. Electroconvulsive therapy also was ineffective. In January 1994, all drugs were withdrawn and clozapine (50 mg four times daily) was started. Dystonia improved during the next 4 weeks. Shortly afterward, however, this therapy was discontinued and replaced with haloperidol decanoate because clozapine was unavailable in Italy and the patient was unwilling to get it from the Vatican pharmacy. The observed improvement rapidly vanished. In April 1994, clozapine was started again and haloperidol was withdrawn. Dystonia rapidly improved during the first 6 weeks of clozapine administration; the clinical improvement further progressed during the following months. The patient is now taking clozapine (250 mg/day) as monotherapy and currently presents with only a mild focal tongue dystonia.

There is no conclusive consensus on the treatment of tardive dystonia. Reduction or withdrawal of neuroleptic drugs, although advisable, is often impractical due to the underlying mental disease. Anticholinergic drugs, tetrabenazine, or reserpine have been reported to alleviate tardive dystonia (2,6). However, based on recent evidence (4,5,7,8) it seems that clozapine is the therapy of choice for tardive dystonia. There are two possible mechanisms by which clozapine may be helpful in tardive dystonia. First this atypical neuroleptic agent may simply allow withdrawal of typical neuroleptic drugs; second, it may be a specific therapy for tardive dystonia. The latter possibility bears some interesting pathophysiologic implications and is supported by the present observation because lingual dystonia rapidly improved when haloperidol was replaced with clozapine and relapsed when clozapine was discontinued; tardive dystonia rapidly improved again when clozapine was administered a second time.

It has been proposed that clozapine is a specific remedy for different movement disorders (9,10); however, its clinical efficacy on idiopathic torsion dystonia (11) and on Huntington's chorea (12) has been disputed. Controlled comparative studies on the efficacy of clozapine in idiopathic and tardive dystonia are warranted in order to assess the clinical efficacy of clozapine and to verify current theories on the pathophysiology of dystonia.

Legends to Videotape

Segment 1. Speech is not intelligible due to lingual dystonia. Simple sensory tricks (e.g., stretching the shoulders, touching the eyes, mouth, or throat) are not helpful. Dystonia does not occur at rest with the face held between the hands, but it reappears as soon as the patient attempts to speak. In order to produce intelligible language, the patient has to bite a handkerchief while speaking. Smoking provides temporary relief, but does not improve language.

Segment 2. The patient has gained weight after clozapine treatment. Language is now clearly intelligible and expeditious. A mild lingual dystonia can still be appreciated. The patient can easily open his eyes and stick his tongue out to a full range.

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