Case Reports

Ziprasidone-induced tardive laryngeal dystonia: a case report

Ching-Shu Tsai, M.D. a, Yu Lee, M.D., M.S. a, Yung-Yee Chang, M.D. b, Pao-Yen Lin, M.D., Ph.D. a,*

Department of Psychiatry, Chang Gung Memorial Hospital-Kaohsiung Medical Center, Chang Gung University College of Medicine, Kaohsiung County 833, Taiwan
Department of Neurology, Chang Gung Memorial Hospital-Kaohsiung Medical Center, Chang Gung University College of Medicine, Kaohsiung County 833, Taiwan

Received 31 May 2007; accepted 22 August 2007

Abstract

Tardive laryngeal dystonia, a rare form of dystonic syndrome, was only reported to be induced by typical antipsychotics. Here, we report one case of ziprasidone-induced tardive laryngeal dystonia in a schizophrenic female patient, who showed dysphonia, hoarseness and dyspnea after taking ziprasidone 120 mg/day for 8 months. These symptoms were significantly improved after discontinuing ziprasidone and increasing the dose of trihexyphenidyl for 1 week. Although atypical antipsychotics are associated with a lower risk of extrapyramidal symptoms, caution should be taken for any tardive dystonic movement when using these medications.

© 2008 Elsevier Inc. All rights reserved.

Keywords: Laryngeal dystonia; Tardive dystonia; Ziprasidone

1. Introduction

Tardive dystonia (TDt) is a neurological syndrome consisting of sustained, involuntary muscular contractions resulting in abnormal postures or repetitive movements. It can be categorized as focal (one body part), segmental (two to four body parts) or generalized (involving face, neck, trunk, at least one upper extremity and at least one lower extremity) [1,2]. Although exposure to antipsychotics is the most common etiologic factor, antiemetics and antidepressants have also been shown to cause TDt [2]. Atypical antipsychotics, such as ziprasidone, have the advantage of a relatively low risk of developing extrapyramidal symptoms. However, there are several case reports of ziprasidone-induced dystonic syndrome, including acute dystonia [3–5], Pisa syndrome [6] and TDt [7]. Tardive laryngeal dystonia, a rare type of TDt, had been found to be associated only with typical antipsychotics [8,9] but not with atypical antipsychotics. Here, we describe a female schizophrenic patient who developed tardive laryngeal dystonia after 8 months of ziprasidone treatment.

2. Case report

A 39-year-old woman, with paranoid schizophrenia, had her initial manifestations of auditory hallucination, delusions of persecution and reference and impaired social and occupational functions at the age of 24 years. She had no history of perinatal or developmental abnormality, general medical disease or drug abuse. From 1992 to 2006, four different antipsychotics have been used, including olanzapine 10 mg/day, sulpiride 600 mg/day, zotepine 75 mg/day and haloperidol 5 mg/day. However, she had irregular drug compliance due to poor insight and intolerable side effects, including extrapyramidal symptoms and weight gain; thus, her psychotic symptoms were exacerbated episodically. Oculogyric crisis had occurred after taking sulpiride 600 mg/day or haloperidol 5 mg/day, separately. In 2006, her regimen was shifted to ziprasidone 80 mg/day and trihexyphenidyl was kept at 6 mg/day to prevent extrapyramidal symptoms. After 2 weeks, ziprasidone was increased up to 120 mg/day due to persistent auditory hallucination with...
reacting emotional response. After being treated with ziprasidone 120 mg/day, her auditory hallucination was rarely noted. Eight months later, she began to have difficulty in phonation and hoarseness, accompanied with shortness of breath. A series of examinations were performed. She had no family history of involuntary movement. No focal neurological deficits or involuntary movements over head, trunk or four limbs were noted during neurological examinations. Physical examination did not reveal any major abnormality, including in the heart and chest regions. Otolaryngological examination excluded the possibility of inflammatory or neoplastic change in the larynx and vocal cord or any traumatic injury and revealed no repetitive abnormal movement in the vocal cords. Blood and biochemical test results were all within normal range, including serum copper and ceruloplasmin levels. Chest X-ray results showed normal findings. Brain computerized tomography showed no special brain pathology except mild cortical atrophy in bilateral parietal areas. Hence, tardive laryngeal dystonia induced by ziprasidone was diagnosed. Her dystonia did not improve after decreasing ziprasidone to 80 mg/day and increasing trihexyphenidyl to 12 mg/day for 1 week. Then, ziprasidone was switched to quetiapine 75 mg/day and trihexyphenidyl was kept at 12 mg/day. One week later, dysphonia and dyspnea were improved significantly, although residual mild difficulty in phonation was observed until 1 month later. One month later, quetiapine was increased to 100 mg/day due to reappearance of auditory hallucination, delusion of persecution and irritability. Trihexyphenidyl was maintained at 12 mg/day to prevent extrapyramidal symptoms. No exacerbation of involuntary movements and improved psychotic symptoms were noted with quetiapine 100 mg/day treatment in the next 5 months.

3. Discussion

In this case, we excluded local laryngeal or chest problem as the cause of difficulty in phonation and hoarseness. Laryngeal dystonia appeared 8 months after continuing ziprasidone treatment and improved significantly after discontinuing ziprasidone. However, we could not exclude the contribution of high-dose trihexyphenidyl in improving dystonia, despite the fact that this effect was not present in the first week after dose increment. Her TDt was diagnosed according to the criteria described by Burke et al. [1], including presence of chronic dystonia, history of preceding antipsychotic drug treatment, exclusion of known causes of secondary dystonia and negative family history for dystonia. Considering the involved region, tardive laryngeal dystonia was impressed. The Naranjo adverse drug reaction probability scale was five in this case, suggesting the involuntary movement was possibly due to ziprasidone. Hence, ziprasidone-induced tardive laryngeal dystonia was diagnosed. However, the possibility of idiopathic dystonia could not be absolutely excluded.

Risk factors of TDt had been identified, such as younger age, male sex, mental retardation and convulsive therapy [10–12], but none was specific to tardive laryngeal dystonia. For ziprasidone-induced TDt, there was only one case reported — a 56-year-old woman with involuntary tongue movement and a problem in jaw opening [7]. Together with the case in current report, both of them are middle-aged females, which are different from the identified risk factors of TDt [10,12]. Because there were only two cases, we could not make any conclusion for the risk factors of ziprasidone-induced TDt.

Although the pathophysiologic mechanism of TDt remains obscure [2], the major focus has been on the dopaminergic and cholinergic mechanisms [13], such as dopamine deficiency and cholinergic excess caused by dopamine antagonists, and on the treatment effect of anticholinergic drugs [14]. The treatment of TDt is composed of discontinuation of the antipsychotic or replacement with a less potent one [15,16] and using anticholinergic agents [16,17]. In our case, we tried both of these modalities. We did not use clozapine in order to avoid the high risk of weight gain and the requirement of frequent blood testing. Significant improvement of her dystonia was observed. Although atypical antipsychotics were reported to be associated with a lower risk of involuntary movement, caution should be taken for any TDt when using these medications.

References


