Spasmodic torticollis associated with sertraline in a child and an adolescent

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Movement disorders or extrapyramidal symptoms (EPS) associated with selective serotonin reuptake inhibitors (SSRIs) have been reported. Although akathisia was found to be the most common EPS, and fluoxetine was implicated in the majority of the adverse reactions, there were also cases with EPS due to sertraline treatment. We present a child and an adolescent who developed torticollis (cervical dystonia) after using sertraline. To our knowledge, the child case is the first such report of sertraline-induced torticollis, and the adolescent case is the third in the literature.

Key words: ***sertraline, torticollis, dystonia, child, adolescent.***

Selective serotonin reuptake inhibitors (SSRIs) have been reported to induce extrapyramidal symptoms (EPS). Several reviews of case reports of EPS ascribed to use of SSRIs found that the most common type of EPS was akathisia, followed by dystonic reactions, parkinsonism and tardive dyskinesia-like states[1]. Fluoxetine was implicated in the majority of the cases of SSRI-induced EPS.

Although fluoxetine has been linked to more cases of EPS in the literature, sertraline, being widely used in children and adolescent psychiatry, has also been known to cause several types of movement disorder[1]. The most common type of EPS associated with sertraline was akathisia, but other EPS types such as dystonia, tremor and reversible choreiform dyskinesia have also been reported[2].

Spasmodic torticollis, also called cervical dystonia, is a kind of movement disorder characterized by abnormal posturing of the neck due to sustained or intermittent muscle contractions[3]. Trauma and drug use are among the most common etiologic factors causing secondary spasmodic torticollis to occur[4].

Here we present two cases of possible sertraline-induced spasmodic torticollis, one in a child and one in an adolescent.

Case Reports

Case 1

A., an 8-year-old boy, was referred to our outpatient unit by his parents due to his excessive worrying about his family, health, school performance and future events. He had also somatic complaints such as headaches and sleep difficulties. These problems had started eight months before referral. In his psychiatric assessment, he was anxious, displayed nail-biting behavior and seemed irritable when worrying about his problems. His intelligence level was within normal limits. His condition was diagnosed as generalized anxiety disorder, for which he had received no previous psychiatric treatment. Sertraline, 25 mg/day, was initiated; the dose was increased to 50 mg/day after one week. By the second visit, four weeks later, his anxiety had decreased and his somatic complaints had ameliorated substantially. We therefore decided to continue the treatment, but two weeks after the second visit, he was admitted to our clinic due to abnormal posturing of his neck and head. Muscle stiffness was detected in his physical examination. There was no significant finding on neurological examination, cranial magnetic resonance imaging and blood work. He had no history of trauma or use of any drug other than sertraline. There were also no indications of

other extrapyramidal signs. We assessed this condition to be torticollis (cervical dystonia), probably induced by sertraline. Sertraline was discontinued, and his symptoms abated with administration of a 5 mg dose of intramuscular biperiden.

Case 2

B., a 16-year-old girl, was admitted to our outpatient unit due to pounding heart, trembling, nausea, shortness of breath and feelings of numbness during the previous two months. She said that her symptoms always began abruptly and diminished after about 30 minutes. She had also concerns about having additional attacks and had had multiple referrals to emergency services in the last two months. She was diagnosed with panic disorder without agoraphobia. Her medical workup was unremarkable; there was no history of psychiatric treatment. Sertraline, 25 mg/day, was initiated; we increased the dose to 50 mg/day after one week. By the second visit, four weeks later, her attacks had ameliorated somewhat, and we thus decided to increase the sertraline dose to 100 mg/day. But three days later, she was referred to our emergency department due to immobility and spasms in the left side of her neck. Painful stiffness was detected in her physical examination. There was no significant finding on neurological examination and blood work. She had no history of trauma or use of any drug other than sertraline. We diagnosed this condition as torticollis (cervical dystonia), probably due to sertraline. A single 5 mg dose of intramuscular biperiden was administered; torticollis resolved within an hour. Sertraline was decreased to 75 mg/day, and on this dosage, the patient experienced neither panic attacks nor side effects.

Discussion

We have reported a child and an adolescent who developed torticollis after sertraline treatment. Extrapyramidal symptoms related to sertraline have been reported in the literature as akathisia, dystonia, tremor and reversible choreiform dyskinesia[2]. Almost all such cases occurred among the adult population. To the best of our knowledge, our case A is the first report of sertraline-induced torticollis in a child, and case B is the third reported adolescent case.

We firstly excluded the possible organic or other conditions that might have caused dystonia in our patients using medical examinations and neuroimaging methods. We also scored the sertraline-induced cervical dystonia in our cases according to the adverse drug reaction probability scale (Naranjo's scale). The adverse reaction observed was classed as "most probably" based on the Naranjo algorithm[5].

There had been only two adolescent cases of dystonia secondary to sertraline use reported in the literature. In the first of these, oromandibular dystonia was reported after the use of sertraline at a dosage of 50 mg/day for a week in a 17-year-old girl[6]. Another adolescent girl was reported to have developed cervical dystonia after the sertraline dose was increased from 50 mg/day to 75 mg/day in the fifth week of the treatment[7]. Intravascular diphenhydramine and intramuscular biperiden, respectively, were applied to overcome dystonia in these two patients. The second of the two resembles our adolescent case in many aspects, such as age, sex and manifestation of the side effect.

However, there was no previous report of sertraline-induced dystonia in childhood. The duration of treatment with SSRIs prior to onset of EPS varied among cases reported in the literature, but was in general relatively short, being only 0-6 days in 33% of the cases[1]. However, in our child case, torticollis due to sertraline use occurred after six weeks without an increase in the dose. In both of our cases, biperiden was effective in treating dystonia, as in adult patients. Studies indicate that extrapyramidal symptoms are more common in females than in males, and that females are more vulnerable to EPS[8,9]. Our child case was, however, a male patient.

The exact mechanism causing SSRI-induced EPS is unknown. Plausible mechanisms include the inhibitory modulation of dopaminergic function in the nigrostriatal pathways; the reciprocal balance between dopaminergic, serotonergic, noradrenergic or cholinergic activity; and central nervous system synaptic potentiation of serotonin and/or norepinephrine through inhibition of transporters or inhibition of their degradation by monoamine oxidase[10-12]. The striatal inhibition of dopaminergic functions by serotonin, the receptor occupancy studies

of serotonin, the receptor polymorphisms and the cytochrome P450 (CYP2D6) phenotypes all lend credence to the various hypotheses presented[13-16]. Female sex, age and pharmacokinetic interaction by CYP2D6 inhibition of concurrent drugs with potential for EPS all appear to be risk factors. The management of SSRI-induced EPS is essentially not different from that of neuroleptic-induced EPS and consists of dosage reduction, discontinuation or change of medication and/or use of anticholinergic drugs, beta blockers, benzodiazepines and, in some cases, dopamine agonists[17,18].

In conclusion, this report aims to increase the clinician's awareness regarding the rare adverse reactions to commonly prescribed drugs that may occur. Moreover, it is not unusual for the pediatric population to develop drug-induced adverse effects. Sertraline, an SSRI, is being widely used in children and adolescents with or without psychiatric comorbidity. Given the widespread use of this drug in this population, uncommon side effects like torticollis should be borne in mind, and clinicians should be aware of such clinical reactions due to psychotropic drugs.

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