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CASE REPORT



A case of an 11-year-old girl diagnosed with OCD developing giggle incontinence following addition of aripiprazole to treatment

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ABSTRACT

Giggle incontinence (GI) is characterized by involuntary and generally unpredictable release of urine during giggling or laughter. It has been suggested that GI may be a centrally mediated disorder and may share a common pathophysiology with narcolepsy and cataplexy. The fact that methylphenidate reduces some symptoms suggests that the condition may be a cataplexy. While alpha 1 and dopamine D2 antagonists exacerbate cataplexy, alpha 1 and dopamine D2 agonists produce a marked improvement. Aripiprazole, frequently used to augment treatment of obsessive compulsive disorder, may have exhibited an alpha 1 and D2 receptor antagonist effect and have caused GI. We describe a case of GI occurring following aripiprazole use in an 11-year-old female obsessive compulsive disorder patient resistant to treatment.

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Giggle incontinence;
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Introduction



Giggle incontinence (GI) is generally seen in girls and is characterized by involuntary and generally unpredictable release of urine during giggling or laughter. The amount of urine released is greater compared to stress incontinence, and the entire bladder is generally evacuated. Patients generally have no lower urinary system symptoms. The most striking characteristic is that the child is usually unable to stop the flow of urine once it has started. This rare disease is quite hard to treat, and its aetiology is still unclear [1]. The fact that methylphenidate reduces some symptoms suggests that the condition may be a cataplexy [2]. Obsessive compulsive disorder (OCD) can severely impair functioning in children and adolescents. Cognitive behavioural therapy (CBT) is the first-line treatment for the mild to moderate OCD. For moderate to severe cases, medication is indicated in addition to CBT. Selective serotonin reuptake inhibitors (SSRIs) are the first choice as drug therapy in OCD. Modification to another SSRI or to clomipramine is recommended in cases in which resistance to one SSRI is observed [3,4].

However, symptoms in most paediatric patients do not resolve entirely with either method. The use of two SSRIs together or of the addition of clomipramine or atypical antipsychotics to an SSRI to which a partial response is obtained are recommended as augmentation therapies [3,4]. Aripiprazole is an atypical

antipsychotic drug that functions by means of partial dopamine and serotonin 1A receptor agonist activity and that has a lower side-effect profile compared with other atypical antipsychotics [5]. Studies involving aripiprazole use in treatment-resistant paediatric OCD cases have been performed [6]. This report discusses a case of GI occurring after addition of aripiprazole to augment treatment in a girl diagnosed with refractory OCD.

Case

An 11-year-old girl, the oldest of three siblings, was living with her parents and two siblings. She was brought to our clinic with symptoms involving frequent hand-washing, repeatedly cleaning her spoon and fork before eating although they were already clean, and irritation resulting from others smacking their lips while eating. Her mother described these symptoms as being present for 3–4 years and worsening in the previous year and reported that the patient had been taken to a child psychiatrist the previous year for that reason and had been started on sertraline. She reported that the initial sertraline dosage of 50 mg had subsequently been augmented to 100 and 150 mg, but that no benefit had been achieved. Sertraline was, therefore, stopped and replaced by anafranil, but anafranil was also discontinued due to side-effects. At interviews, the patient reported washing her hands 25–30 times a day and

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constantly regarding everything as dirty, and that she was irritated by people around her constantly smacking their lips as they ate. We were informed that she frequently became involved in arguments over these obsessions at home and at school. At examination, she was appropriate for her age with low frustration tolerance. Her history was unremarkable. In terms of family history, we learned that her mother had been treated for OCD and that her maternal aunt had a childhood history of GI.

Following interviews and evaluations, the patient was diagnosed with OCD on the basis of DSM-5 criteria, and cognitive behavioural therapy was applied. The clinical global impression–severity of illness (CGI-SI) score was 6 and the Children’s Yale-Brown Obsessive Compulsive Scale (CY-BOCS) score was 26 (severe). A CBT protocol involving one weekly 45-min session for 16 weeks was planned. The patient was first given exposure and response prevention behavioural techniques directed towards the compulsion producing the least anxiety (washing her fork and knives before eating) as homework and started on 10 mg fluoxetine. The patient did not attend the weekly therapy sessions, and when she did arrive after 3 weeks, she had not done her homework. We learned there had been little decrease in her obsessions and that the irritation persisted. On the patient’s second visit to our clinic, her clinical global impression–global improvement (CGI-GI) score was 3.

Fluoxetine was raised to 20 mg, and we strongly recommended that the homework be performed. When the patient returned for check-up a further 2 weeks later, no significant decrease occurred in her symptoms, and fluoxetine was incrementally raised to 60 mg. On her presentation, she had again not done her homework, and she was, therefore, assessed as therapy non-compliant, and CBT was discontinued. At subsequent follow-ups, we learned that the cleanliness obsessions had partially decreased, that the obsession with lip smacking persisted, but that there had been a decrease in irritability. On the patient’s visit to our clinic, her CY-BOCS score was 15 (mild), her CGI-SI score 3, and her CGI-GI score 2. Aripiprazole 5 mg was, therefore, added to the treatment (initiated at 2 mg/day and gradually titrated up to 5 mg/day over one week). The patient was brought back to our clinic after 2 weeks. We learned that sudden incontinence had occurred while laughing in school 2 days after taking aripiprazole, that she had been unable to retain the urine during incontinence, and that when this recurred several times, she had presented to the urology clinic. All tests performed there were normal, and we were informed the patient had been diagnosed with GI. The family reported that this phenomenon had occurred after the newly initiated medication, and the urologist suggested seeking the advice of a psychiatrist. Aripiprazole was then stopped, and the

patient was invited back for follow-up 2 weeks later. At that subsequent interview, we learned there had been no recurrence of urinary incontinence following discontinuation of aripiprazole.

Discussion

We describe a female patient with OCD developing GI following aripiprazole use. The incontinence resolved entirely following discontinuation of aripiprazole. Her Naranjo adverse drug reaction probability scale score for the drug was 6 [7]. GI in children is rare but easily identified by history. The typical patient has incontinence associated only with laughter. The characteristic feature of GI is sudden urinary incontinence beginning with giggling, and total voiding of the bladder in association with an inability to halt the flow of urine. No urinary system pathology has been determined in several GI patients, and treatment is difficult. This is due to a failure to identify the underlying aetiological factors [8]. GI may develop as a result of central nervous system-mediated activation of the pontine micturition centre. It has been suggested that GI may be a centrally directed disorder and may share a common pathophysiology with cataplexy and narcolepsy. Cataplexy, one of the determining symptoms of narcolepsy, is characterized by a sudden decrease in muscle control stimulated by emotions, and particularly laughter [9]. Pharmacological research has shown that preventing norepinephrine intake mediates the anti-cataplectic effects of currently prescribed antidepressants, while stimulating dopamine intake and/or dopamine secretion mediates the stimulating effect of central nervous system stimulants [10]. Although the role of dopaminergic activation varies in the brain, it has been suggested that GI and cataplexy may share a common pathophysiology [8]. Alpha 1 receptors play an important role in cataplexy control. Alpha 1 antagonists exacerbate cataplexy, while alpha 1 agonists significantly improve it. Alpha 1 receptor agonists and central dopamine D2 agonists suppress cataplexy [11,12].

Aripiprazole is a novel antipsychotic agent with a different effect mechanism to both typical and atypical antipsychotics. In contrast to other atypical antipsychotics, aripiprazole exhibits a partial agonist effect on dopamine D2 receptors and an antagonistic effect on serotonin 5-HT_{2A} receptors. In addition, aripiprazole can exhibit antagonistic effects despite exhibiting low affinity to adrenergic alpha 1 receptors [13]. It is well-tolerated. The most commonly reported side-effects, with an incidence of 5% or more compared to placebos, are insomnia, tremor, akathisia, nausea, and vomiting. These reported side-effects are generally mild or moderate and tend to disappear after the first week of treatment. However, Parkinson’s disease, Parkinsonism, akathisia, dyskinesia, acute dystonic reaction, and neuroleptic malignant syndrome triggered by

aripiprazole have appeared in the form of rare case reports. Despite being a partial D2 agonist, aripiprazole has been shown to cause side-effects such as parkinsonism by behaving like a D2 antagonist [14]. There are also case reports of aripiprazole causing priapism by exhibiting antagonistic effects on alpha 1 receptors [15]. In the light of these details, GI being a variant of cataplexy and the antagonistic effect of aripiprazole on alpha 1 and D2 receptors exacerbating cataplexy, suggested that the drug may have triggered GI. However, much is still unknown, such as the exact aetiology of GI and the mechanism by which aripiprazole triggers it. This case suggests that not using aripiprazole as the first therapeutic option when antipsychotics are required in patients with a family history of GI can both reduce the risk of side-effects and also increase compliance with treatment. Further studies are now needed to clarify the aetiology of GI and the mechanism by which drugs exhibit positive or adverse effects.

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Disclosure statement

No potential conflict of interest was reported by the authors.

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