A case of the giggles

Diagnosis and management of giggle incontinence

Lisa Fernandes Pharmd RPh Danielle Martin MD CCFP FCFP MPubPol Susan Hum MSc

iggle incontinence (GI) is an unusual condition of involuntary total bladder emptying triggered by laughing or giggling.1 Giggle incontinence can be difficult to recognize, as embarrassment can prevent disclosure of symptoms, and it is difficult to treat. Although it is much more common in girls, we describe a case of GI in an adolescent boy.

Case

A 14-year-old boy presented to an urban academic family practice health centre with concerns of total bladder emptying when he laughed, no matter where he was. His incontinence started at a young age, but had worsened recently. It occurred about 3 times per week, only with laughing, and not with coughing, sneezing, or running. He was becoming increasingly anxious about having embarrassing accidents at school and during extracurricular activities (ie, hockey and baseball), and contemplated avoiding school and sport activities.

The patient's medical history was unremarkable. He had no known drug allergies and did not take any

medications. He had potty trained easily as a child. He had experienced constipation when he was 3 to 4 years old, but not since that time. He reported no polydipsia, no polyuria, and no nocturnal enuresis. On examination, he looked well. His growth was normal and he was not overweight. His abdomen was soft and nontender, with no masses and no organomegaly. His bladder was not palpable. His genitalia were normal, with an uncircumcised penis, an easily retractable foreskin, and a normal urethral orifice. His pubertal development was appropriate at Tanner stage 3. Investigation results did not contribute to a diagnosis: urinalysis results were normal and his serum glucose reading was 5.7 mmol/L. His family physician recommended Kegel exercises and timed voiding.

He returned to the clinic 2 months later with no improvement. The physician consulted a local pediatric urologist by telephone who diagnosed him with GI. A trial of 10 mg of oral methylphenidate (MPH) twice a day (MPH is usually prescribed for attention deficit hyperactivity disorder) was recommended.

Editor's key points

- Giggle incontinence is a relatively rare form of daytime wetting characterized by involuntary, unstoppable, complete bladder emptying during or immediately after laughing or giggling. Giggle incontinence is distinct from stress incontinence, in which a small amount of urine leaks with sneezing, coughing, or straining.
- It appears that encouragement and reassurance of cure, combined with behaviour modification, is the best first-line approach for many patients. From a family medicine perspective, this nonpharmacologic approach is appropriate, as primary care providers might need to counsel and help their patients with issues of low self-esteem, social isolation, or anxiety.
- Regardless of giggle incontinence severity, first-line treatment options include reassurance of cure and timed voiding or bowel management, followed by biofeedback and Kegel exercises. Second-line treatment of oxybutynin with timed voiding could be tried, followed by third-line treatment with methylphenidate.

Points de repère du rédacteur

- L'incontinence au rire est une forme relativement rare de perte urinaire diurne qui se caractérise par le vidage involontaire, impossible à arrêter et complet de la vessie durant ou immédiatement après un épisode de rire ou de fou rire. Elle se distingue de l'incontinence à l'effort qui prend la forme de fuites d'urine en petites quantités lors d'un éternuement, de la toux ou d'un effort.
- Il semble que pour de nombreux patients, le fait de les encourager et de les rassurer à propos de la guérison, combiné à des modifications de comportements, représente l'approche à privilégier. Dans le contexte de la médecine familiale, cette stratégie non pharmacologique est appropriée, car les médecins de soins primaires pourraient être appelés à conseiller et à aider leurs patients à propos de problèmes comme une faible estime de soi, l'isolement social ou l'anxiété.
- Qu'importe la gravité de l'incontinence au rire, les options thérapeutiques de première intention incluent le fait de rassurer quant à la guérison, et la miction à intervalles fixes ou la prise en charge de la fonction intestinale, suivis par la rétroaction biologique et les exercices de Kegel. On peut essayer un traitement de deuxième intention comportant de l'oxybutynine de concert avec la miction à moments fixes, puis un traitement de troisième intention avec du méthylphénidate.

The team pharmacist counseled the patient to take the first MPH dose before school and the second dose 4 to 5 hours later to ensure coverage near the end of the school day. Methylphenidate could be omitted on weekends, as his symptoms would be less of an issue and he would have easier access to a washroom. Common side effects, such as mood, sleep, and appetite changes, were reviewed.

In a telephone follow-up 9 months later, the mother stated that MPH had helped, with no reported side effects. She believed the GI symptoms were either related to a growth spurt or the intensity of extracurricular activities near the end of the school term. She remarked that both her mother and her aunt had had similar experiences when they were younger. His current GI symptoms were "not considered an issue anymore" and he discontinued MPH. There were no subsequent visits for GI.

Discussion

Giggle incontinence, also known as giggle micturition or enuresis risoria, is a relatively rare form of daytime wetting characterized by involuntary, unstoppable, complete bladder emptying during or immediately after laughing or giggling. Bladder function is normal at other times.1

Giggle incontinence is distinct from stress incontinence, in which a small amount of urine leaks with sneezing, coughing, or straining.2 The pathogenesis of GI is unclear, although it is postulated to be centrally mediated and related to a receptor imbalance of cholinergic and monoaminergic systems. Giggle incontinence and cataplexy might share a common pathophysiology, as laughter or emotions might trigger muscle hypotonia.3 Giggle incontinence mostly affects early or mid-pubescent girls, and is potentially a hereditary disorder associated with a strong female family history of this syndrome.4 The natural history of untreated GI and whether medications shorten its course is not known or well documented.4

An Ovid MEDLINE, PubMed, and PubMed Central literature search (1996 to week 4 of January 2016) was conducted with the terms giggle incontinence, enuresis risoria, and treatment. Studies of nonpharmacologic interventions (biofeedback and Kegel exercises) and drug treatment (oxybutynin plus timed voiding and MPH) were described.

Biofeedback with Kegel exercises was evaluated in children who did not respond to timed voiding or to treatment with anticholinergic agents or pseudoephedrine. Two-thirds of children who attended 4 or more weekly or biweekly biofeedback sessions had a full response that endured at least 6 months; those who attended fewer sessions achieved only a partial response.5

Oxybutynin (0.2 to 5 mg/kg twice a day) with timed voiding was evaluated in 109 children (70% girls). After 10 weeks, wetting frequency was improved and total remission was achieved in 89% of patients.6

In 3 studies that evaluated the effectiveness of MPH, most of the children (70% to 90% female; mean age ranging from 11 to 16 years) had a strong family history of GI; wetting frequency ranged from once monthly to multiple events daily. Treatment included varying daily oral doses (0.2 to 0.5 mg/kg or 5 mg) and formulations (intermediate or short-acting) of MPH. Treatment duration ranged from 2 months to more than 5 years.^{3,7,8} Decreased appetite (n=2) and difficulty falling asleep (n=1) were reported in 1 study. Giggle incontinence symptom resolution took up to 5 years. Therefore, it remains unclear whether patients were cured or simply outgrew GI. A recent retrospective study reported that daily GI frequency and dysfunctional voiding are predictive factors of treatment resistance, and one treatment option, 5 mg MPH daily plus behavioural urotherapy for at least 6 months, had a maximum success rate of 56%.9

In 1981, the presentation and management of GI were described in 2 preadolescent boys with a 1- to 2-year history of symptoms and a wetting frequency of 4 to 5 times a week. At the outset, both boys were reassured that they could be cured. The 13-year-old had GI only when he was standing, and usually when he was with friends. He was advised to sit down if he started to laugh, and to take 15 mg of propantheline (a parasympatholytic drug) 1 hour before going out with friends. After 6 months, a placebo was substituted and within 3 months, he was continent. The 11-year-old had a very strong family history of enuresis. For 3 weeks, he recorded in a diary each episode of wetting and the reasons for laughing. During that time, he only had 3 days of slight wetting of his pants. In the subsequent 3 weeks, he had no wetting episodes, and was confident that he was cured.10

Based on the above-mentioned studies, it appears that encouragement and reassurance of cure, 10 combined with behaviour modification (and using incontinence pads or underpants for backup protection), is the best first-line approach for many patients. From a family medicine perspective, this nonpharmacologic approach is appropriate, as primary care providers might need to counsel and help their patients with issues of low self-esteem, social isolation, or anxiety. Anecdotally, bloggers have reported suffering in silence (too embarrassed to discuss GI symptoms with doctors, parents, or teachers), feeling "like a baby," and feeling anxious or embarrassed for having wetting accidents; many were bullied by their peers. As it is still unclear whether medications actually cure GI, second- and third-line treatment with oxybutynin and MPH, respectively, should be considered a last resort.

Conclusion

Clinicians might not be aware of or recognize GI, because many children and preadolescents might be too embarrassed to disclose their symptoms. Giggle incontinence is also difficult to treat. However, regardless of GI severity,

first-line treatment options could include reassurance of cure and timed voiding or bowel management, followed by biofeedback and Kegel exercises. Second-line treatment of oxybutynin with timed voiding could be tried, followed by third-line treatment with MPH. It should be noted that the latter option might be objectionable to many parents owing to its association with attention deficit hyperactivity disorder.

Dr Fernandes is a clinical pharmacist at the Family Practice Health Centre at Women's College Hospital in Toronto, Ont. Dr Martin is a family physician and Vice President for Medical Affairs and Health System Solutions at Women's College Hospital and Associate Professor in the Department of Family and Community Medicine and the Institute of Health Policy, Management and Evaluation at the University of Toronto. Ms Hum is Research Associate in the Department of Family and Community Medicine at Women's College Hospital.

Competing interests

None declared

Correspondence

Dr Lisa Fernandes; e-mail lisa.fernandes@wchospital.ca

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