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CASE REPORTS

A case of wet laughs

Um caso de risos molhados

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ABSTRACT

Introduction: Giggle incontinence (GI) is a rare form of daytime urinary incontinence characterized by involuntary, unstoppable, and large-volume voids with complete bladder emptying during or immediately after laughing.

Case report: A 15-year-old girl presented to the Family Health Unit with concerns of involuntary bladder leakage while laughing, sometimes with total bladder emptying, during the past two years. Symptoms did not manifest while coughing, sneezing, straining, or with nocturnal enuresis. No complaints of dysuria, pollakiuria, or constipation were reported. On physical examination, the girl showed adequate growth and weight, and urine analysis, urine culture, and renal-vesical ultrasound were normal. She was referred to the Pediatric Outpatient Clinic of a secondary hospital, where GI was diagnosed.

Discussion: GI is a clinical syndrome with potentially underestimated incidence. Due to their closer relationship with patients, family doctors should be aware of this condition and recognize its symptoms.

Keywords: adolescent; daytime urinary incontinence; enuresis; giggle incontinence

RESUMO

Introdução: A incontinência risória (IR) é uma forma rara de incontinência urinária diurna, caracterizada por perdas involuntárias incontroláveis de grandes volumes urinários, com esvaziamento vesical completo durante ou imediatamente após o riso.

Caso clínico: Uma adolescente de 15 anos recorreu à Unidade de Saúde Familiar com queixas de perdas urinárias involuntárias com o riso, com esvaziamento vesical completo, desde há dois anos. Negava perdas involuntárias com a tosse, espirro, ou esforço ou enurese noturna e não apresentava sintomas de disúria, poliaquiúria ou obstipação. Ao exame físico, apresentou crescimento e peso adequados, bem como sedimento urinário, urocultura e ecografia renovesical normais. A rapariga foi referenciada para a consulta de Pediatria de um hospital secundário, onde foi diagnosticada IR.

Discussão: A IR é uma síndrome clínica com incidência potencialmente subestimada. Devido à relação mais próxima que mantêm com os doentes, os médicos de família devem estar alerta para esta condição e reconhecer a sua sintomatologia.

Palavras-chave: adolescente; enurese; incontinência do riso; incontinência urinária diurna

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INTRODUCTION

According to the Committee of the International Children's Continence Society terminology, giggle incontinence (GI), or enuresis risorii, is a relatively rare form of daytime incontinence, which refers to urine leakage occurring only with laughter.[1] It is characterized by involuntary, unstoppable, large-volume voids, with complete bladder emptying during or immediately after laughing.[1,2] It should be distinguished from stress incontinence, where there is a small amount of urine leakage when sneezing, coughing, or straining. On the other hand, bladder function is usually normal when children affected are not laughing.[1,2] The condition should be differentiated from others, like overactive bladder, voiding postponement, or underactive bladder, in which children may experience urine leakage during sudden lapses of concentration, including laughter.[1]

Regarding epidemiology, GI is more frequent in females and may be associated with a family history of the condition.[3] It predominantly affects early or mid-pubescent girls and often resolves spontaneously in puberty, although some cases persist into adulthood.[3]

The pathogenesis of GI is uncertain. Many authors support the theory that GI is centrally mediated by an undefined neurological cascade that follows laughter. It has even been hypothesized that GI might have a functional relationship with cataplexy, since laughter or emotion may trigger muscle atonia in the pelvic floor, leading to urinary incontinence.[2,4] Other authors defend that detrusor instability can also contribute to the disease, concluding that GI is a combination of neurologic and urologic factors.[4]

The diagnosis of GI is usually established based on the clinical history and supported by absence of other voiding symptoms and normal complementary means of diagnosis.[5] Constipation is occasionally reported by patients, although the mechanism by which it may be associated with GI is still poorly understood.[6] Delayed diagnosis has also been reported, as patients feel embarrassed to reveal their symptoms.[2]

Currently, GI management has shown only limited success. Based on the hypotheses about the pathogenesis of the condition, available treatments consist of behavioral measures (including lifestyle changes and bladder/pelvic floor retraining with biofeedback techniques) and pharmacotherapy (including methylphenidate, anticholinergics, antidepressants, and anticonvulsants).[3,7] However, a standard treatment is yet to be established.

CASE REPORT

Herein is reported the case of a 15-year-old girl living with her parents. Family APGAR score revealed a highly functional family, with medium Graffar class and in Duvall's cycle phase VI.

The girl was observed at a Family Health Unit consultation accompanied by her mother, with concerns of involuntary bladder leakage only while laughing, with two years of evolution. The volume of outflow was not constant, varying from large-volume (with total bladder emptying) to low-volume leakage. Episodes occurred about two to three times a week at school, home, or wherever the trigger occurred. The girl denied involuntary urinary loss while coughing, sneezing, straining, or with nocturnal enuresis. Other symptoms of vesical dysfunction were excluded, including alterations in daytime voiding frequency, urinary urgency, difficulty in emptying the bladder, straining, or urine flow pattern changes. No complaints of dysuria or pollakiuria were reported, as well as no constipation.

The patient was previously healthy, with adequate stature-ponderal progression and no history of recurrent urinary tract infections or reno-vesical pathology before or after birth. According to the mother, she acquired daytime sphincter control around the age of 24 months and nocturnal control at three years. She did not take medications regularly and had no drug allergies documented. The family history was also unremarkable, with absence of reno-vesical pathology.

During physical examination, the patient only presented a slightly tympanitic abdomen. The external genitalia and additional examinations were normal, as well as pubertal development (Tanner stage 5).

Complementary diagnostic tests including urinalysis, urine culture, and reno-vesical ultrasound were performed, showing no alterations. However, post-void residual urine volume was not assessed.

One month after the first Family Health Unit consultation, the patient was observed at the Pediatrics Outpatient clinic of a secondary hospital, where GI diagnosis was established. The patient and parents were assured of the benign nature of the syndrome. Behavioral measures with lifestyle changes were proposed as treatment strategy. However, the patient missed the following hospital consultation and was lost to follow-up. Two years later, she was observed at the Family Health Unit for a non-related issue and reported spontaneous symptom resolution.

DISCUSSION

GI is a well-defined clinical syndrome characterized by urine leakage occurring specifically during or immediately after laughing. The incidence of the condition may be underestimated because patients underreport symptoms due to embarrassment, on the one hand, and because patients, families, and even health professionals fail to recognize symptoms, on the other.[2] Patients sometimes report low self-esteem, social isolation, anxiety, and depressive symptoms due to GI.[2] Support from family and friends is crucial for patients to preserve self-esteem and maintain quality of life.[6]

The incidence of GI is higher in women, a fact that can be explained by recent evidence showing that men and women activate different parts of the brain when responding to humor.[8]

In this case report, the patient only searched for a doctor two years after symptom onset, which clearly delayed the diagnosis. Although GI is a potentially hereditary disorder and frequently associated with

positive family history, there were no reports of GI on other family members in the present case. The absence of relevant findings on physical examination and normal complementary diagnostic test results were compatible with GI, which is a diagnosis of exclusion.

Before implementing specific treatment measures, it is important to establish GI diagnosis and explain the benign nature of the syndrome to patients and their families since the prognosis is usually favorable, with spontaneous symptom resolution, and only a few cases persist into adulthood.[6] Treatment is not properly established yet, with a wide range of approaches mainly involving behavioral and pharmacological measures used. Behavioral measures include lifestyle changes and biofeedback exercises. It seems that the best first-line approach for many patients includes lifestyle measures with timed voiding, correct position during micturition or defecation (both feet on a flat surface and coordination between relaxation of pelvic floor muscles and bladder contraction), bowel management to prevent constipation, adequate fluid intake during the day, and restricted consumption of caffeine, chocolate, and citrus fruits.[2,9] Biofeedback techniques are used for pelvic floor retraining, to correct dysfunctional voiding patterns and associated complications. Children and adolescents are taught to identify the external sphincter muscles and control them more rapidly and forcefully to decrease incontinence.[3] In one small case series, the biofeedback technique was effective in nine patients either refractory to pharmacotherapy or whose parents refused medication, reason why it might be recommended before pharmacotherapy.[3]

Regarding GI pharmacological treatment, disparate results have been obtained in studies with anticholinergics, antidepressants, anticonvulsants, and methylphenidate, the latter being the only agent showing some promise in this setting.[3] Some authors advocate the use of anticholinergics based on the assumption that GI is related to detrusor instability.[10] Suggesting a central nervous system etiology, Berry *et al.* conducted a trial of methylphenidate in patients who met GI criteria and accepted to take the medication. Results showed that incontinence episodes during schooltime completely resolved in 12 of the 15 patients, although three patients reported side effects, like decreased appetite and delayed sleep onset.[8] Others side effects have also been commonly reported, like stomach ache or headache, jitteriness, and social isolation, being more pronounced with higher medication doses.[8] Methylphenidate is not yet approved by the Food and Drug Administration, being only used off-label at this time.[7]

In conclusion, family doctors should be aware of GI symptoms, taking advantage of juvenile and infantile health surveillance appointments to actively ask patients about enuresis and vesical dysfunction. The close relationship of these physicians with their patients may be useful since many children and adolescents may be too embarrassed to disclose their symptoms. A behavioral approach is appropriate in Family Medicine setting, and counseling can be helpful in assuring patients of the benign nature of this syndrome and helping them cope with associated problems, like low self-esteem, anxiety, and depression.

AUTHORSHIP

Patrícia Graça Silva - Conceptualization; Writing – original draft; Writing – review & editing

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