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Air Swallowing Caused Recurrent Ileus in Tourette's Syndrome

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Abstract

This report describes an adolescent boy who has Tourette's syndrome and developed a subtle but significant increase in vocal tics after an 8-month respite. The increase in vocal tics was associated with an acute increase in psychological stressors and resulted in recurrent air swallowing, which, in turn, caused abdominal cramping, eructation, and flatus, eventually leading to aeroenteria. Air swallowing was recognized only after a second hospital admission for recurrent ileus. Air swallowing and associated symptoms were mitigated by reinstitution of psychopharmacologic treatment and an increase in the patient's self-awareness of the air-swallowing behavior. Clinically significant air swallowing has not been described previously in Tourette syndrome or a tic disorder. This case is important for pediatricians and pediatric gastroenterologists because either may be the first to evaluate a child or an adolescent with unexplained recurrent ileus. This report also documents the importance of the connection between the brain and the body.

Keywords

abdominal pain; aerophagia; air swallowing; Tourette's syndrome; ileus

Tourette's syndrome (TS), once thought to be a rare disorder, may affect up to 1.5% of young adolescents.¹ This disorder is characterized by the presence of both vocal and motor tics, not necessarily occurring concomitantly, for at least 1 year.² This disorder typically presents in childhood with repetitive eye blinking or other repetitive stereotyped ocular, facial, or shoulder movements.³ Although vocal tics can manifest as sudden repetitive vocalization of noises, barks, syllables, words, or phrases, it is not uncommon for vocal tics to be much more subtle, manifesting as episodes of sudden repetitive sniffing, throat clearing, grunting, or swallowing. The diagnosis of TS can be difficult because tics usually wax and wane in intensity and severity, with occasional periods of prolonged quiescence, and because motor tics usually precede vocal tics by several years. Morbidity that is associated with TS uncommonly is attributable to the tics themselves but is usually a result of comorbid aggression, attention-deficit/hyperactivity disorder, obsessive-compulsive disorder (OCD), learning disabilities, anxiety, and/or depression.^{2,4}

Excessive air swallowing can cause abdominal distention, cramping, colic, flatulence, and eructation. In such cases, radiologic studies may demonstrate large volumes of air in the stomach and intestine.⁵ Prolonged air swallowing can result in a gastrointestinal disorder known as aerophagia. Although aerophagia can be caused by neurologic disorders, anatomic abnormalities of the trachea or pharynx, or chronic obstructive respiratory disease, functional

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behaviors, such as talking excessively while eating and habitual air swallowing, also can result in aerophagia. Severe aerophagia can cause ileus, volvulus, or bowel necrosis.⁶ Treatment for aerophagia typically consists of behavioral modification therapy.⁷

The current report describes an adolescent who had TS and who, after 8 months without tics, experienced a subtle but significant increase in vocal tics after an acute increase in psychological stressors. Air swallowing that resulted from the tics eventually led to unexplained recurrent ileus. The diagnosis was not made until the second hospital admission and then only after the second examination by the consultation team. Although the consequences of air swallowing are discussed in the adult medical literature, a review of the major textbooks of pediatrics is notable for few discussions regarding this entity. Furthermore, although eructation has been associated with habitual repetitive air swallowing, termed “eructio nervosa,”⁸ clinically significant symptoms of air swallows have never been documented in TS until this report. This case is of the utmost importance to pediatricians, pediatric gastroenterologists, and pediatric surgeons because any may be the first to evaluate a patient who has unexplained recurrent ileus that may present as a surgical abdomen. This case also is an example of the intimate relationship between the brain and the body.

CASE REPORT

A 16-year-old boy developed eye blinking and throat clearing at 7 years of age and started psychopharmacologic treatment for TS at 11 years of age. Initial treatment with citalopram and quetiapine was unsuccessful. Subsequently, the combination of risperidone 0.5 mg twice a day and fluoxetine 40 mg once a day controlled his symptoms for ~2 years.

Because of a cessation of his symptoms, risperidone was discontinued and fluoxetine was tapered to 30 mg once a day. Approximately 8 months later, the patient began to experience several psychological stressors, including losing his summer job, final examinations, and becoming aware of his father’s diagnosis of leukemia. During a 2-month period, the patient experienced excessive flatulence, eructation, and, occasionally, large explosive bowel movements. Elimination of beans and dairy products from his diet did not relieve gastrointestinal symptoms. The patient denied nausea, vomiting, diarrhea, dyspepsia, hematochezia, melena, steatorrhea, sick contacts, fever, rash, or weight loss during this period. In retrospect, during this time, the patient noticed an increase in eye blinking, throat clearing, and swallowing.

The patient developed intermittent abdominal cramping associated with eructation during an out-of-state trip to attend his grandmother’s funeral. These symptoms increased in intensity and frequency during a 2-day period. The severity of the abdominal pain resulted in admission to an outside hospital. Complete blood count and electrolytes were unremarkable. The patient received a diagnosis of having an ileus and was treated with nasogastric tube placement. His symptoms improved, he passed a bowel movement, and he was discharged the next morning.

During the 7-hour automobile trip back to his home city, the patient’s abdominal cramping and eructation increased, prompting evaluation in the emergency department (ED). In the ED, the patient’s vital signs were stable. He was alert and oriented with normal cognition but seemed anxious. Belching and flatus was obvious. His head, ears, eye, neck and throat, cardiovascular, pulmonary, extremity, skin, and neurologic examinations were normal. The abdomen was nontender, mildly distended, and diffusely tympanic with bowel sounds. There was no stool in the rectal vault, and guaiac was negative. Complete blood count, electrolytes, calcium, magnesium, phosphorous, serum urea nitrogen, creatinine, alkaline phosphatase, albumin, total protein, aspartate transaminase, alanine aminotransferase, amylase, lipase, and total and direct

bilirubin were normal. Supine and upright abdominal films demonstrated aeroenteria extending from the stomach to the rectum without air-fluid levels (Fig 1).

The patient had a history of mild speech delay, mild intermittent asthma, mild eczema, anxiety, and depression. The patient irregularly received interpersonal therapy from a psychiatrist for ~3 years. The patient's mother has dopamine-responsive dystonia and had a history of speech delay, and his father was in remission from leukemia. The patient's mother, father, and maternal and paternal grandparents experienced intermittent rage. The patient's maternal grandmother had mixed collagen vascular disease. There was no family history of Crohn's disease, ulcerative colitis, irritable bowel syndrome, or celiac spruce.

The patient was admitted and instructed to refrain from liquid or solid intake. Intravenous fluids and ranitidine 50 mg 4 times per day was provided. Frequent air swallowing was not noticed by the ED or by the primary care team but was recognized by the gastroenterologic consult service only after the second examination. Antiendomysial, transglutaminase, antigliadin, and total IgA all were normal. Immunoglobulin G for *Helicobacter pylori* was negative. Because the patient was able to tolerate liquids and solids without abdominal pain or nausea and had a formed bowel movement, he was discharged 2 days after admission. At discharge, omeprazole 20 mg once a day was added to the patient's admission psychopharmacologic treatment.

Immediately after discharge, fluoxetine was increased to 40 mg/day, and risperidone was reinstituted at 0.25 mg in the morning and 0.5 mg at bedtime. Three weeks after his admission, the patient's tics had decreased substantially and his gastrointestinal symptoms did not recur. Omeprazole was discontinued at that time. Risperidone was discontinued 1 year after admission, and tics remained quiescent 2 years after admission. The patient currently is being treated for a mood disorder and anxiety with venlafaxine. The patient admits to feeling "little swallow" during times of increase stress. With awareness of this symptom, the patient has been actively able to suppress repetitive swallowing and prevent recurrences of abdominal symptoms.

DISCUSSION

This report describes a 16-year-old boy who has TS and developed moderately severe air swallowing during an 8-week period. The patient's acute increase in vocal tics paralleled his gastrointestinal symptoms. The patient's symptoms were relieved with a short course of bowel rest, reinstitution of psychopharmacologic treatment, and an increase in self-awareness of the process that caused his abdominal symptoms. This is the first report of clinically significant air swallowing associated with TS.

Air swallowing has been reported in several neurologic and psychiatric syndromes. Aerophagia has been reported to cause ileus, volvulus, or bowel necrosis in children with mental retardation⁶ and airway obstruction in schizophrenia.⁹ Aerophagia has been associated with the abnormal movements of Huntington's disease¹⁰ and the abnormal breathing pattern of Rett syndrome¹¹ but not with repetitive stereotyped movements in non-degenerative neurologic disorders such as a tic disorder or TS.

Incessant eructation has been reported as the presenting symptom in OCD.¹² Premonitory midline abdominal and throat sensations have been associated with a compulsive urge to perform tics in TS.¹³ Therefore, compulsive repetitive stereotyped movements, such as vocal tics or repetitive swallow, not uncommonly seen in either OCD or TS, may be derived from common neurobiological mechanisms. If voluntary or involuntary repetitive vocal tics alone were responsible for the excessive air swallowing in the current patient, then an association between air swallowing or aerophagia and TS or OCD would be expected to be better known. The patient in this report had some unique characteristics that may have contributed to the

development of his symptoms. For example, acute decomposition of the patient's stable state with a rapid increase in the frequency of his repetitive swallowing could have prevented the development of equilibrium between the entering of air into the bowel through vocal tics and typical routes for gas release. In addition, the patient's anxiety most likely increased as his gastrointestinal symptoms increased, resulting in an increase in his TS symptoms, more vocal tics, and, therefore, more aeroenteria.

The differential diagnosis of aerophagia includes mal-absorption syndromes, anatomic obstruction, infection, bacterial overgrowth syndrome, intestinal pseudo-obstruction, cystic fibrosis, and celiac disease.¹⁴ However, the subacute onset and rapid resolution of this patient's symptoms make such conditions unlikely. Although fluoxetine and risperidone have been associated with stomach pain and constipation, this patient was stable on these medications for >1 year, and the onset of this patient's symptoms corresponded to a reduction in the dose of these medications. The diagnosis of aerophagia requires symptoms to occur for at least 12 consecutive or nonconsecutive weeks within a 12-month period.¹⁴ Although the patient described in this case did not reach the criterion for aerophagia because his symptoms lasted only 8 weeks, the air swallowing that he experienced resulted in moderately severe complications that required 2 hospitalizations.

This case illustrates a new association between a medical disorder and severe air swallowing. Because tic disorders and idiopathic aerophagia have a similar age of onset, the former should be considered in the differential diagnosis of causes of the latter in children and adolescents. Behavioral therapy can be effective at mitigating the symptoms of either disorder, but in severe cases, at least for TS and tic disorders, pharmacologic treatment usually is necessary.

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Abbreviations

TS	Tourette's syndrome
OCD	obsessive-compulsive disorder
ED	emergency department

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FIGURE 1. Supine (A) and upright (B) abdominal plain films demonstrating aeroenteria extending throughout the gastrointestinal system.