High Cost and Low Yield: The Diagnostic Evaluation of Rumination Syndrome in Pediatrics

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Objective To document the use of diagnostic testing in adolescents who ultimately were diagnosed with rumination syndrome, a functional gastrointestinal disorder. We examined the diagnostic yield of each test as well as the associated costs, and we determined if any demographic or illness-related variables impacted the magnitude of the work-up.

Study design A retrospective chart review was conducted for 68 patients with rumination syndrome admitted to our inpatient treatment program. The cost and findings of patients' diagnostic investigations were gathered, as well as demographic and illness-related variables to determine factors that may be related to evaluation size.

Results The most commonly used tests in the evaluation of rumination syndrome included esophagogastroduodenoscopy, gastric emptying, antroduodenal manometry, upper gastrointestinal series, and abdominal ultrasound scan. Each patient underwent an average of 8.8 tests, with the average cost for each patient's diagnostic work-up being US $19 795. Few tests were found to be beneficial in the diagnosis of rumination syndrome, and few demographic or illness variables were found to be related to the overall extent of the investigation.

Conclusions Extensive testing for rumination syndrome in adolescents is common in clinical practice, and comes at a high financial cost with low yield, likely delaying diagnosis and treatment. Symptom-based criteria should be used to make the diagnosis of rumination syndrome. *(J Pediatr 2017;185:155-9).*

Rumination syndrome is a functional gastrointestinal disorder characterized by recurrent, effortless regurgitation or expulsion of food or fluids that begins soon after ingestion[1] (**Table I**). Variants of rumination have been documented across the lifespan and in children across the spectrum of cognitive abilities.[2]

The diagnosis can be made by obtaining a detailed clinical history and conducting an observation of the rumination.[3-5] Even so, there often is a considerable lag between the commencement of rumination symptoms and the final diagnosis.[6] This may occur for several reasons, including the practitioner's lack of familiarity with the disorder, discomfort with making the diagnosis of a functional gastrointestinal disorder, and/or the overlap in symptoms between rumination syndrome and other gastrointestinal conditions (eg, gastroesophageal reflux disease, achalasia, gastroparesis) or clinical eating disorders (eg, bulimia nervosa).

Prior studies have documented the plethora of diagnostic tests that patients ultimately diagnosed with rumination syndrome undergo. Chial et al[7] found that, in their sample of 147 children and adolescents with rumination syndrome, the average number of diagnostic tests was 3.3 (range 0-8). The most common tests included an upper gastrointestinal (UGI) series, upper endoscopy (esophagogastroduodenoscopy [EGD]), gastric emptying, brain imaging, and antroduodenal (AD) manometry. The authors emphasized that none of the tests were particularly useful in making the diagnosis of rumination syndrome, that rumination itself can impact the test findings (eg, a gastric emptying study showing a delay in emptying not because of gastroparesis, but because of the regurgitation itself), and that such a significant work-up is quite costly and at times misleading to families.

Although these findings were informative and provided rich data about patients with rumination, there were several limitations. First, the sample of patients was diverse in age (range 5-20 years) and included several patients with developmental disabilities. Second, as the study was a chart review, the authors were dependent upon the search finding patients with a diagnosis of rumination. Finally, the chart review was conducted on patients seen over the course of a 25-year period, resulting in a heterogeneous sample.

The current study was conducted to document common practices in the use of diagnostic testing in a group of patients who were diagnosed with rumination syndrome under Rome III criteria; to examine the positive yield of each test; to estimate the costs associated with these work-ups; and to determine whether or

AD	Antroduodenal
API-4	Abdominal Pain Index-4
CSI-24	Children's Somatization Inventory-24
EGD	Esophagogastroduodenoscopy
UGI	Upper gastrointestinal

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Table I. Rome III diagnostic criteria for adolescent rumination syndrome

Criteria fulfilled at least once per week for at least 2 months before diagnosis, and must include all of the following:

1. Repeated painless regurgitation and rechewing or expulsion of food that
   a. begin soon after ingestion of a meal
   b. do not occur during sleep
   c. do not respond to standard treatment for gastroesophageal reflux
2. No retching
3. No evidence of an inflammatory, anatomic, metabolic, or neoplastic process that explains the subject's symptoms

not any demographic or illness-related variables impacted the extent of the work-up. We hypothesized that the amount of testing would be greater for patients with a longer duration of rumination symptoms, greater somatic complaints, reduced health-related quality of life, and those requiring special nutritional support because of the severity of their rumination.

Methods

A chart review was conducted for the first consecutive 74 patients admitted to our inpatient treatment program from 2009 to 2015. Insufficient data were available for 6 patients, and these patients were removed from the sample, resulting in a total sample of 68 adolescents. Each patient met Rome III criteria for rumination syndrome.[1] Patients were predominantly female (86.8%), similar to that seen in other samples of adolescents with rumination syndrome. Fifty percent of patients required special nutritional support (eg, gastric or jejunal tube feedings or total parenteral nutrition) at the time of their admission to our program. There was considerable variability in the duration of time patients had symptoms of rumination syndrome prior to receiving treatment at our center (range = 3-163 months, median = 21 months). Other patient characteristics are described in **Table II**. Many of the patients in the current chart review were described in a prior study by our group.[8]

Diagnostic Study Data

Because the majority of patients were referred from outside hospitals, diagnostic test data were extracted from electronically shared medical records and information forwarded by outside physicians as part of the initial referral. For internal referrals from our institution, data were obtained directly from the electronic medical record. We examined demographic information, medical history (eg, the current use of enteral or parental feeding), and past diagnostic testing (ie, radiologic, endoscopic, and motility). The only diagnostic tests considered for the current study were (1) those that explore UGI functioning and (2) those that took place between the dates of rumination symptom onset and admission to our program.

Table II. Patient characteristics and questionnaire scores

Variables	N	Mean (SD)
Age (y)	68	15.8 (2.3)
CSI-24 (parent)	30	1.3 (0.5)
CSI-24 (patient)	44	1.5 (0.7)
API-4 (parent)	36	2.3 (1.2)
API-4 (patient)	46	2.0 (1.2)
PedsQL 4.0-Total (parent)	30	64.9 (18.5)
PedsQL 4.0-Total (patient)	45	63.9 (18.3)

PedsQL, Pediatric Quality of Life Inventory.

Cost Data

Information regarding the cost of the diagnostic investigations was gathered from 2 sources. First, for each investigation, data regarding the "national pricing" of the test was obtained via the Pediatric Health Information System. The Pediatric Health Information System is a comparative pediatric database that contains pricing information for each test (as determined by the test's current procedural terminology code) across 45 children's hospitals. For the current study, the "national pricing" component of the cost was the average pricing for the investigation across comparison hospitals. Second, regarding the "professional charge" for each investigation (eg, test interpretation), national comparison data were not available. Therefore, the "professional charge" component of the total cost was obtained through our own hospital system for each investigation.

Taken together, the "total cost" data (**Table III**) was the sum of the national pricing for the diagnostic test and the professional charge associated with that individual test. The "grand sum" for each patient's complete diagnostic work-up consisted of the total cost across all radiologic, endoscopic, and motility tests the individual patient received.

Somatic Symptoms and Abdominal Pain

To index the presence and severity of somatic symptoms, parents and patients completed the Children's Somatization Inventory-24 (CSI-24) and the Abdominal Pain Index-4 (API-4).[9] The CSI-24 is a well-validated measure that asks patients (and parent by proxy report) about 24 nonspecific somatic complaints that may have occurred over the previous 2 weeks. Participants were asked to describe how much they were "bothered" by each symptom in the previous 2 weeks, with responses ranging from 0 (not at all) to 4 (a whole lot). Parents were asked to report on their impressions of their child's somatic symptoms. The most common method of scoring the CSI-24 entails calculating a total score, which is the average of the scores across all 24 items (with a maximum score of 4.0). Higher scores represent greater symptom presence. This total score has been shown to have good internal consistency ($\alpha = 0.88$). The CSI-24 was not used from our program's inception, therefore, only 30 parent and 44 patient questionnaires were available for use in the current study (**Table II**).

The API-4[10] is a 4-item measure on which the individual (or parent) rates the frequency, duration, and intensity of the patient's abdominal pain over the past 2 weeks. In line with the scoring procedure advocated by the authors,[11] all items were converted to a 5-point scale, and a composite score created (with a maximum score of 4.0). The authors validated the measure