

Novel association of rectal evacuation disorder and rumination syndrome: Diagnosis, comorbidities, and treatment

United European Gastroenterology Journal 2014, Vol. 2(1) 38-46 © Author(s) 2014 Reprints and permissions: sagepub.co.uk/journalsPermissions.nav DOI: 10.1177/2050640613518774 ueg.sagepub.com



Priya Vijayvargiya*, Johanna Iturrino*, Michael Camilleri, Andrea Shin, Maria Vazquez-Roque, David A Katzka, Jill R Snuggerud and Richard J Seime

Abstract

Background: Patients with disorders of gastrointestinal function may undergo unnecessary treatment if misdiagnosed as motility disorders.

Objective: To report on clinical features, medical, surgical, and psychiatric comorbidities, and prior treatments of a patient cohort diagnosed concurrently with nonpsychogenic rumination syndrome and pelvic floor dysfunction (also termed rectal evacuation disorder).

Methods: From a consecutive series (1994–2013) of 438 outpatients with rectal evacuation disorders in the practice of a single gastroenterologist at a tertiary care centre, 57 adolescents or adults were diagnosed with concomitant rumination syndrome. All underwent formal psychological assessment or completed validated questionnaires.

Results: All 57 patients (95% female) fulfilled Rome III criteria for rumination syndrome; rectal evacuation disorder was confirmed by testing of anal sphincter pressures and rectal balloon evacuation. Prior to diagnosis, most patients underwent multiple medical and surgical treatments (gastrostomy, gastric fundoplication, other gastric surgery, ileostomy, colectomy) for their symptoms. Psychological comorbidity was identified in 93% of patients. Patients were managed predominantly with psychological and behavioural approaches: diaphragmatic breathing for rumination and biofeedback retraining for pelvic floor dysfunction.

Conclusions: Awareness of concomitant rectal evacuation disorder and rumination syndrome and prompt identification of psychological comorbidity are keys to instituting behavioural and psychological methods to avoid unnecessary treatment.

Keywords

Psychological comorbidities, rectal evacuation disorder, rumination syndrome

Received: 4 October 2013; accepted: 5 December 2013

Introduction

Based on questionnaire studies, upper gastrointestinal symptoms such as reflux, heartburn, and postprandial fullness are experienced by up to one-third of adults in the USA. Constipation is reported in 15% adults and 20–25% in elderly or African American adults. There is overlap between functional gastrointestinal disorders (FGID), such as functional dyspepsia and irritable bowel syndrome (IBS), and gastrooesophageal reflux and IBS. Therefore, these symptom combinations may be presenting complaints in adults attending primary care or gastroenterology clinics. These symptom complexes may be attributed to visceral hypersensitivity or impaired gastrointestinal motility and are treated

with limited success. There is no treatment proven to benefit patients with overlapping syndromes.

On the other hand, rumination syndrome and rectal evacuation disorders are forms of upper and lower FGID that are characterized by more-specific symptom profiles or objective test findings. Rumination

Mayo Clinic, Rochester, USA

*loint first authors

Corresponding author:

Michael Camilleri, Clinical Enteric Neuroscience Translational and Epidemiological Research (CENTER), Mayo Clinic, Charlton 8-110, 200 First St. SW, Rochester, MN 55905, USA.

Email: camilleri.michael@mayo.edu

Vijavvargiva et al. 39

Table 1. Required and supportive criteria of rumination in 57 patients with rectal evacuation disorder-rumination

Criteria	Rome III criterion for rumination	Patients (%)
Required	Persistent or recurrent regurgita- tion of recently ingested food into the mouth with subse- quent spitting or remastication and swallowing	100
	Regurgitation is not preceded by retching	100
Supportive	Regurgitation events are usually not preceded by nausea	39
	Cessation of the process when the regurgitated material becomes acidic	7
	Regurgitant contains recognizable food with a pleasant taste	61
	Daily or almost daily	95
	Every meal or almost every meal	89

Values are %. Criteria fulfilled for the last 3 months with symptom onset at least 6 months before diagnosis.

syndrome is characterized by almost daily, repeated, effortless regurgitation of partially digested food typically within 30 min of meal ingestion, with subsequent rechewing and reswallowing or expulsion;^{3,4} diagnosis is based on the Rome III criteria⁵ (Table 1). Rumination has been described in children, adolescents, and adults of normal intelligence.^{3,6} Less than 20% of patients with rumination syndrome have a prior history of anorexia or bulimia nervosa; however, these are usually resolved at the time of presentation with rumination.

Pelvic floor dyssynergia or rectal evacuation disorders (RED) cause obstruction to defecation, and are characterized by incomplete relaxation of the pelvic floor and external anal sphincter, resulting in constipation. RED are diagnosed by findings on rectal examination (such as limited perineal descent, high anal sphincter tone at rest, and tenderness or paradoxical contraction of the pelvic floor during attempts to simulate defecation) and anorectal manometry and balloon expulsion test. In our referral practice, RED were the cause of chronic constipation in >25% patients in a consecutive series of >1400 patients.8,9 Rumination and RED are treated with behavioural approaches, 10 with success rates approximating 70% in patients with RED. Similarly, in our practice, 15/214 patients referred for evaluation of upper gastrointestinal symptoms had rumination syndrome.¹¹

Our study objective was to report on a patient cohort diagnosed concurrently with both rumination syndrome and RED in order to gain insight into potential diagnostic cues or disease associations that will aid clinicians to diagnose and manage the disorders.

Methods

Search strategy in electronic medical records

We utilized the proprietary Data Discovery and Query Builder at the Mayo Clinic, Rochester to enter the search queries 'pelvic floor dysfunction', 'pelvic floor dyssynergia', 'evacuation disorder', 'obstructive defecation', 'outlet obstruction', 'dyssynergic defecation', and 'Camilleri'. We further narrowed the results by searching for 'rumination syndrome', 'regurgitation', and 'vomiting'.

Participants

In a retrospective study of electronic medical records between 1 January 1994 and 30 April 2013, we identified a cohort of 438 patients with RED, among whom 57 (13%) were diagnosed with concomitant rumination syndrome.

The Mayo Clinic Institutional Review Board approved this study for patients who had consented to use their medical records for research.

Diagnostic criteria

Rumination syndrome was determined based on the required Rome III criteria (Table 1), including two additional supportive criteria (D and E): occurrence daily or almost daily; and involving every meal or almost every meal.⁵

RED was suspected on clinical symptoms that included features consistent with Rome III criteria for functional defecation disorders (i.e. symptoms consistent with functional constipation), ¹² as well as objective findings. Thus, RED was confirmed on balloon expulsion test (requiring >200 g of weight) and maximum resting anal sphincter pressure >90 mmHg on anorectal manometry. Additional criteria, based on scintigraphic defecography, were failure of the anorectal angle to open >15° with straining, or perineal descent >4.5 cm. ¹³ These criteria were based on a review of the literature and appropriate age ranges. ^{8,14,15}

Data extraction

Data extracted included demographics, clinical features, medications, past medical history, social history, physical examination, evaluation, comorbidities, surgeries, treatments, and outcome.

Gastrointestinal transit studies

Gastric emptying was measured by following a standardized breakfast meal tagged with ^{99 m}Tc sulphur colloid via gamma camera images taken immediately postprandially and at 1, 2, and 4 h. For colonic transit measurement, patients ingested ¹¹¹In adsorbed on activated charcoal delivered to the colon by a methacrylate-coated, delayed-release capsule taken with the standardized breakfast for gastric transit. Abdominal scinticans were obtained at 8, 24, and 48 h after ingestion of the ¹¹¹In capsule. ^{16,17}

Other tests

We also recorded other tests conducted at Mayo Clinic and medically relevant ones performed at other institutions. These included measurements of gastric accommodation by single-photon-emission computed tomography, ¹⁸ oesophageal manometry, oesophageal pH studies, endoscopy and other imaging studies.

Psychological assessment

Psychological diagnoses and personality characteristics were based on the patients' previous psychiatry history, clinical interview by behavioural therapist and psychologist (Masters and Doctoral level psychology team), and validated psychological questionnaires (including Generalized Anxiety Disorder 7 [GAD-7] and Patient Health Questionnaire [PHQ-9]). 19–22

Analysis

Descriptive data are displayed as number of patients (%) or mean ± standard error of the mean. Proportions of findings are based on the numbers of patients in whom the specific parameter was documented in the record.

Results

Participant characteristics

Of the 438 patients evaluated for RED over the 19 years under study, 57 (13%) were identified with concomitant rumination syndrome. Age at diagnosis was (mean \pm standard deviation) 30.3 ± 1.63 years (range 14–62, eight <18 years) and body mass index was $20.8\pm0.5\,\mathrm{kg/m^2}$ (range 14.8–32.8). Fifteen patients (26%) were considered underweight (<18.5 kg/m²). In this patient group, 95% were female and 89% were Caucasian.

Table 2. Clinical features suggestive of rectal evacuation disorder

Clinical feature	Patients	
RED history		
Support perineum	2/26 (5)	
Anal digitations	7/35 (20)	
Vaginal digitations	0/28 (0)	
Excessive strain	32/40 (80)	
Incomplete rectal evacuation	15/22 (68)	
Digital rectal examination ^a		
Decreased perianal sensation	0/38 (0)	
Decreased perineal descent (≤1 cm)	43/53 (81)	
High resting anal sphincter pressure	26/52 (50)	
Paradoxical contractions	24/37 (65)	
Puborectalis tenderness	28/39 (72)	
Rectal examination combinations		
All	9/56 (16)	
Three	12/56 (21)	
Two	17/56 (30)	
One	15/56 (27)	
None	3/56 (5)	

Values are n with feature present/n with feature documented in medical record (%).

Clinical features: findings in the history

The primary complaints were vomiting or regurgitation in 28% of the study population, constipation in 14%, and both in 58% (n = 57).

Rumination. All 57 participants fulfilled both of the required and supportive Rome III criteria for rumination syndrome (Table 1).

Rectal evacuation disorder. Fifty-one (89%) of the 57 patients reported symptoms of constipation. Although they did not specifically complain of constipation, detailed history showed that five of six patients had history suggesting a disorder of rectal evacuation, specifically two with sense of incomplete rectal evacuation, one supporting the perineum to defecate, pressure on the lower abdomen to facilitate defecation, and five with excessive straining to defecate. In these six patients, routine digital rectal examination identified a nonrelaxing pelvic floor or other features that were suggestive of rectal evacuation disorder and led to further investigation. Table 2 shows the number of participants who endorsed a history of manoeuvres to evacuate stool: perineal support, vaginal or anal digitations, or sense of incomplete rectal evacuation, findings consistent with RED.

^aOne patient who did not undergo digital rectal examination for religious reasons, did undergo anorectal manometry.

Vijavvargiva et al. 41

Onset of RED and rumination. In five patients (9%), rumination had preceded constipation for between 3 months and 10 years. Five patients (9%) noticed feeling constipated once the rumination started. In 23 patients (40%), constipation presented first, followed by rumination (range of months to >10 years). In the remaining 17 patients (30%), there was not enough information to determine the relative time of onset of the rumination and constipation. Six patients (11%) denied feeling constipated. Thus, among the 34 patients with sufficient documentation, constipation preceded rumination in 23/34 (68%) of patients.

Physical examination findings

The findings on rectal examination are summarized in Table 2, which details abnormal perianal sensation, perineal descent, resting anal sphincter tone, paradoxical sphincter contraction upon attempted defecation, and puborectalis tenderness. The most common finding was decreased perineal descent (≤1 cm) observed in

81% (43/53 with this finding recorded) of patients. Patients presented with different combinations of the rectal examination findings, with 57% of patients manifesting one or two characteristics of RED.

Findings on gastrointestinal evaluation

Table 3 summarizes tests that were performed for evaluation of upper and lower gastrointestinal symptoms. Results of scintigraphic assessment of gastrointestinal and colonic transit were available in $\sim 70\%$ of patients (Tables 3 and 4) and compared to controls in our laboratory. ^{16,23} Six patients had accelerated gastric emptying at 2 h (all normal at 4 h), and seven patients had delayed gastric emptying at 4 h (2 being borderline slow). Seven patients had decreased gastric accommodation after a standard 300 ml liquid nutrient (Ensure, 1 kcal/ml) drink (normal postprandial volume >428 ml).

All 57 patients underwent anorectal manometry and balloon expulsion testing; 11 patients also had

Table 3. Investigations to evaluate upper and lower gastrointestinal symptoms

Evaluation	Patients
Rumination	
Gastric emptying	
2 h	
Slow, <25% emptied	4/45 (9)
Rapid, >79% emptied	6/45 (13)
4 h	
Slow, <76% emptied	7/45 (16)
Decreased gastric accommodation (<428 ml postprandial-fasting volume)	7/12 (58)
Abnormal oesophagogastroduodenoscopy	1/15 (7) (reflux oesophagitis)
Oesophageal manometry	1/11 (9) (nutcracker oesophagus)
Oesophagogram	0/6 (0)
Prolonged oesophageal pH monitor	0/3 (0)
Rectal evacuation disorder	
Anorectal manometry $+$ balloon expulsion	
Added weight (g) balloon (>200 g)	45/55 (82)
Maximum resting sphincter pressure (>90 mmHg)	28/57 (49)
Anorectal angle (<15°)	6/11 (55)
Ballooning perineum (>4.5 cm)	0/11 (0)
Decreased perineal descent (\leq 1 cm)	5/11 (45)
Colonic transit	
24 h	
Slow	5/40 (13)
Rapid	4/40 (10)
48 h	
Slow	3/19 (16)

Values are n positive results/n available results (%). Gastric emptying normal values are based on values from 319 healthy controls.²²

anorectal angle and perineal descent measurements as part of the scintigraphic defecography test, which was conducted at our institution until 2008 (Table 4). Four patients had a normal anorectal manometry; in two patients, the test had been conducted at an outside institution which did not evaluate all of the aforementioned parameters. One patient had ingested two types of oral benzodiazepines before the anorectal manometry, but had highly suggestive history and examination findings of RED. Twenty-eight patients (49%) had only one abnormality and 24 participants (42%) had ≥2 abnormalities on tests for RED.

Comorbidities and surgery experienced prior to identification of the combined RED and rumination

A summary of the medical and surgical comorbidities identified prior to the diagnosis of RED-rumination syndrome are summarized in Table 5. Thus, prior to the diagnosis of RED-rumination syndrome, the medical records review showed that 26 (46%) patients had undergone prior gastrointestinal surgeries: 17 cholecystectomies, seven partial or total colectomies, 11 multiple surgeries including percutaneous endoscopic jejunostomy placement. Similarly, the records documented that, prior to the diagnosis of RED-rumination syndrome, 18 patients (32%) had problems with urinary bladder emptying.

Eighteen patients (30% of the 54 in whom this information was available) had to leave college, graduate school, or work, requiring the aid of disability insurance, and even more individuals had to restrict their social or physical activities because of their illness.

Prior medical and nutritional treatments

Fifty-two patients (91%) were prescribed medications $(2.5 \pm 0.2 \, \text{medications})$ for either of their

gastrointestinal presentations and were referred because of unsatisfactory symptomatic relief. Eight patients (14%) were taking both antidepressants and anxiolytics; 17 patients (30%) both antiemetics and antacids; and nine patients (16%) both laxatives and prokinetic agents.

Sixteen patients (28%) required enteral or parenteral nutrition. Eleven patients received enteral nutrition through percutaneous endoscopic jejunostomy and eight patients received parenteral nutrition. Three patients who had started with percutaneous endoscopic

Table 5. History of comorbidities and prior treatments for the combined presentation with rumination and rectal evacuation disorder

Comorbidities	Patients
Medical	
Bladder voiding issues	18/57 (32)
Medications	
Antidepressants	16/57 (28)
Anxiolytics	16/57 (28)
Narcotics	9/57 (16)
Antiemetics	27/57 (47)
Acid blocker (PPI/H2B)	32/57 (56)
Laxatives	25/57 (44)
Prokinetic agents	18/57 (32)
Surgical	26/57 (46)
Cholecystectomy	17/57 (30)
Colectomy	7/57 (12)
Appendectomy	6/57 (11)
Gastric electrical stimulation	2/57 (4)
Fundoplication	1/57 (2)
>1 surgery	11/57 (19)

Values are n positive history/n available history (%).

Table 4. Scintigraphic transit and rectal evacuation

Physiological measurement	Patients (n = 57)	Normal values
Gastric emptying at 2 h (%)	$\textbf{52.5} \pm \textbf{3.6}$	50 (25-78.5)
Gastric emptying at 4 h (%)	$\textbf{86.0} \pm \textbf{2.5}$	96 (76.2-100)
Colonic transit (GC 24 h)	2.3 ± 0.2	2.3 (1.3-4.4)
Colonic transit (GC 48 h)	$\textbf{3.0} \pm \textbf{0.3}$	3.8 (1.9-5.0)
Average resting anal sphincter pressure (mmHg)	$\textbf{91.5} \pm \textbf{5.2}$	<90
Balloon expulsion test (g)	435 ± 28	<200
Perineal descent (cm)	1.7 ± 0.4	>1.5
Anorectal angle change from rest to defecation (°)	11.1 ± 3.9	>15

Values are mean \pm standard error of the mean, median (5–95th percentiles), or cut-off value. GC, geometric centre.

Vijavvarajva et al. 43

jejunostomy continued regurgitating and were eventually placed on parenteral supplementation.

Psychiatric comorbidities prospectively identified at the time of identification of RED and rumination syndrome

Fifty-four patients completed the psychological assessment, including the questionnaires GAD-7 and PHQ-9, which are summarized in Table 6 based on the diagnoses provided by professional psychologists (JRS, RJS). Fifty (93%) had at least one psychiatric comorbidity. The most common psychiatric comorbidity. The most common psychiatric comorbidities were prior history of anorexia or bulimia nervosa, abuse, anxiety, depression, and somatization disorder. Thirty-three patients (61%) were diagnosed with a major disorder based on Diagnostic and Statistical Manual of Mental Disorders, fourth edition.²⁴

Management instituted for RED-rumination

Patients received standard care for both rumination and RED. Rumination behavioural therapy was provided as one session with a behavioural psychologist with instruction on the use of diaphragmatic breathing to abort or control the regurgitation of food. All patients were referred to a biofeedback programme for RED; 32 pursued treatment elsewhere. Twenty-five patients underwent intensive therapy for RED at

Table 6. History of psychiatric comorbidities for the combined presentation with rumination and rectal evacuation disorder

Psychiatric comorbidities	Patients
Anorexia nervosa	7/57 (12)
Bulimia nervosa	3/57 (5)
Abuse (emotional, physical, or sexual)	18/54 (33)
Depression	24/54 (44)
Diagnosis	11/54 (20)
Questionnaire	12/54 (22)
Previous treatment	1/54 (2)
Anxiety	20/54 (37)
Diagnosis	13/54 (24)
Questionnaire	4/54 (7)
Previous treatment	3/54 (6)
Somatization disorder ^a	12/54 (22)
Adjustment disorder ^a	3/54 (6)
Bipolar disorder type I	1/54 (2)
Two psychiatric diagnoses	7/54 (13)
Type A personality characteristics	26/54 (48)

Values are n positive history/n available history (%).

the Mayo Clinic, consisting of multiple sessions of pelvic floor and anal sphincter relaxation with biofeed-back cues provided by electromyography recordings from a plug placed in the anal sphincter. Of these, all except three patients had a resolution of their symptoms; however, they were all subsequently diagnosed as slow colonic transit after resolution of RED and underwent colectomy.

Discussion

Our medical records study has identified a hithertounrecognized association of RED with rumination syndrome in 57 patients. When both conditions were observed in the same patient, 93% of patients had a psychiatric disorder, as compared to 17% reported in rumination⁶ and 65% in 60 patients with RED alone, among patients evaluated at the same tertiary referral centre.²⁵ It is unclear whether the comorbid psychiatric disorders represent the cause or the consequence of RED-rumination. Certainly, the number of failed prior therapies. including enteral and parenteral nutrition, percutaneous gastrostomies, cholecystectomy, and colectomy, may influence the development of secondary psychiatric comorbidities.

The association between FGID and psychiatric diagnoses has been widely explored. For example, in a study of 175 patients with IBS by Rome III criteria (median age, 41 years; 78% women, which were similar to the age and gender in our patient cohort) evaluated at two specialty care clinics, 47% had at least one comorbid mental illness, with the most common classes being anxiety disorders (69%) and affective disorders (38%, of which 22% were depressive disorders.²⁶ In our study, 93% (50/54) of patients had a psychiatric diagnosis, elevated scores on validated psychiatric questionnaires (GAD-7 and PHQ-9), or underwent treatment for a psychiatric disorder. Thus, the proportion of patients with the RED-rumination association with psychiatric disorders appears higher than that observed in IBS patients in tertiary care clinics²⁶ or in patients with rumination alone (66%) in a small prospective study²⁷ and 17% in a larger retrospective analysis of 147 children and adolescents. Because of the unknown prevalence of rumination syndrome in the community, it can be challenging to determine the influence of psychiatric disorders on this condition. However, Green et al.²⁸ have shown that, when psychiatric disorders are present, it is helpful to address these issues for successful treatment, indicating a substantial role of the psychiatric disorder in rumination syndrome.

The observation of 93% psychiatric comorbidity in the RED-rumination cohort is also higher than the 47% of patients with RED in whom there were associated axis-I psychiatric disorders, more commonly

^aPsychiatric diagnosis by psychologist.

anxiety and phobic-anxious temperament, in one study²⁹ and depression, obsessive compulsiveness, anxiety, and high somatization symptom scores in another study.³⁰

Our data show that the majority of psychiatric diagnoses in patients with RED-rumination association were depression (44%), anxiety (37%), or somatoform disorders (22%), consistent with previously reported data in patients with either rumination or RED.^{6,30} Similarly, 48% of our study participants displayed type A personality characteristics, including high grade point average, multiple extracurricular activities. and academic/career achievements.⁶ Studies have shown that abuse (physical, sexual, or psychological) is associated with increasing prevalence of rumination and levator ani syndrome as compared to controls. Furthermore, 23% of female patients with pelvic floor complaints disclosed prior sexual abuse.31 In our current cohort of patients, we found 32% had a history of physical, psychological, or sexual abuse, 18% had either anorexia or bulimia, and 13% (7/54) had some indication of secondary gain from their rumination syndrome such as desired weight loss. These aspects of the patients' histories may provide important clues for accurate diagnosis of RED-rumination. A recent report from Australia also identified an association between pelvic floor dysfunction and more significant eating disorders in 184 patients requiring hospitalization for the eating disorder.³² Overall, our observations suggest that the two conditions and their concurrence represent behavioural disorders, but we are unable to attribute the aetiology to any particular cause such as an abnormal response to visceral sensations or the history of abuse identified in 32% of our cohort.

This patient population was noted to have multiple and severe comorbidities, with many permanent effects secondary to medical interventions. Ninety-one per cent of our patients were prescribed medications, with an average of 2.5 ± 0.2 prescriptions per person. Although these medications may have provided partial short-term relief, none targeted the underlying pathophysiology and, thus, could not provide a long-term solution. Surgical intervention was conducted in 44% of patients, including 17 cholecystectomies and seven colectomies. Sixteen people (28%) were unable to maintain their nutrition and required enteral or parenteral supplementation. Even with the predominance of overachieving individuals (30% had to leave college, graduate school, or work, requiring the aid of disability), even more individuals had to restrict their social or physical activities.

Identification of the RED-rumination association and the extremely high prevalence of the psychiatric comorbidity are highly relevant to providing correct management and avoiding unnecessary treatments and iatrogenic disease. Concurrence of upper and lower gastrointestinal symptoms such as regurgitation, vomiting, bloating, and constipation is not uncommon and may suggest a spectrum of disorders from IBS to more life-threatening, diffuse neuromyopathic diseases. Diagnosis of these neuromyopathic diseases requires extensive, often costly evaluations and may lead to nutritional, medical, or surgical treatments including enteral or parenteral nutrition, percutaneous gastrostomy, fundoplication, ileostomy, or colectomy, as was observed in our tertiary referral cohort. We believe that it is important, from risk, cost, and optimized management perspectives, to identify a less severe association that combines such upper and lower gastrointestinal symptoms, especially since it is diagnosed by simple clinical and laboratory methods and is treated noninvasively.

Our experience suggests that the presenting symptoms of RED-rumination association reflect the components of each syndrome individually. However, it is essential to ask about both regurgitation and constipation symptoms in each individual presenting with either symptom, since the information is often not volunteered at initial evaluation. Six of the 57 patients presenting with rumination denied a history of constipation; however, a rectal examination and anorectal manometry showed classical features of RED.

Behavioural therapy and biofeedback have been effective treatments for the vast majority of patients with rumination syndrome⁶ and RED.³³ The most important predictor of success for physical therapy is a willingness to comply with treatment protocol.³⁴ In patients with psychiatric disorders, lack of motivation and an ideology of failure may interfere with the treatments, based on behavioural modification. Thus, a thorough psychiatric evaluation should be performed in patients with RED-rumination and, in addition to the behavioural and physical therapy to help improve symptom resolution, the required psychiatric therapy should also be provided. Because some psychiatric disorders may be secondary to RED and rumination syndrome,³⁵ proper treatment of RED-rumination may result in an improvement or resolution of the psychiatric comorbidities.

Limitations of this study include the identification of 57 patients in a retrospective medical record review, the reliance on validated psychiatric questionnaires, and a single psychological evaluation by a clinical psychology team who also provided the education and behavioural treatment. However, we lack outcome data on the behavioural treatment, a weakness that is inherent in the retrospective medical records review. The true prevalence of the condition cannot be estimated. RED has been identified in 27% of constipation patients⁸ and rumination syndrome in 15 of 214

Vijavvargiva et al. 45

patients referred for evaluation of upper gastrointestinal symptoms¹¹ at the same tertiary referral centre. A final word of caution is that a minority of patients may have a more complex pathophysiology to explain the occurrence of constipation; thus, three of these 57 patients eventually underwent colectomy after rehabilitation of the RED was insufficient to resolve their constipation due to concomitant slow colonic transit constipation.

The identification of this novel association is justified by the significant medical, social, and academic comorbidities noted and by the unnecessary tests and treatments undergone by patients with RED-rumination. Prospective, longitudinal studies will be beneficial to further characterize this combined disorder, determine the natural history and outcomes of both RED and rumination syndrome, and clarify effective treatment options, including the option of treating exclusively the psychiatric disorder and assessing the outcome of the two gastrointestinal disorders. Future studies will also need to explore the possible pathophysiological mechanisms underpinning the association between rumination and RED. The roles of the diaphragm and the pelvic floor in the pathophysiology of the two conditions suggest that excessive contraction or failed relaxation are worthy of further exploration, including the influence of psychological stress and anxiety.

Funding

This research was supported by the National Institutes of Health (grants DK67071 and DK92179 to MC).

Conflict of interest

The authors declare that there is no conflict of interest.

References

- Camilleri M, Dubois D, Coulie B, et al. Prevalence and socioeconomic impact of upper gastrointestinal disorders in the U.S.: results of the U.S. upper gastrointestinal study. *Clin Gastroenterol Hepatol* 2005; 3: 543–552.
- Stewart WF, Liberman JN, Sandler RS, et al. Epidemiology of constipation (EPOC) study in the United States: relation of clinical subtypes to sociodemographic features. Am J Gastroenterol 1999; 94: 3530–3540.
- O'Brien MD, Bruce BK and Camilleri M. The rumination syndrome: clinical features rather than manometric diagnosis. *Gastroenterology* 1995; 108: 1024–1029.
- 4. Chial HJ and Camilleri M. A twenty-one-year-old college student with postprandial regurgitation and weight loss. *Clin Gastroenterol Hepatol* 2006; 4: 1314–1317.
- Tack J, Talley NJ, Camilleri M, et al. Functional gastroduodenal disorders. *Gastroenterology* 2006; 130: 1466–1479.
- Chial HJ, Camilleri M, Williams DE, et al. Rumination syndrome in children and adolescents: diagnosis, treatment, and prognosis. *Pediatrics* 2003; 111: 158–162.

 Lembo A and Camilleri M. Chronic constipation. N Engl J Med 2003; 349: 1360–1368.

- 8. Nullens S, Nelsen T, Camilleri M, et al. Regional colon transit in patients with dys-synergic defaecation or slow transit in patients with constipation. *Gut* 2012; 61: 1132–1139.
- Shahid S, Ramzan Z, Maurer AH, et al. Chronic idiopathic constipation: more than a simple colonic transit disorder. J Clin Gastroenterol 2012; 46: 150–154.
- Wagaman JR, Williams DE and Camilleri M. Behavioral intervention for the treatment of rumination. *J Pediatr* Gastroenterol Nutr 1998; 27: 596–598.
- Bredenoord AJ, Chial HJ, Camilleri M, et al. Gastric accommodation and emptying in evaluation of patients with upper gastrointestinal symptoms. *Clin Gastroenterol Hepatol* 2003: 1: 264–272.
- 12. Bharucha AE, Wald A, Enck P, et al. Functional anorectal disorders. *Gastroenterology* 2006; 130: 1510–1518.
- 13. Pezim ME, Pemberton JH, Levin KE, et al. Parameters of anorectal and colonic motility in health and in severe constipation. *Dis Colon Rectum* 1993; 36: 484–491.
- Rao SS, Hatfield R, Soffer E, et al. Manometric tests of anorectal function in healthy adults. *Am J Gastroenterol* 1999; 94: 773–783.
- Fox JC, Fletcher JG, Zinsmeister AR, et al. Effect of aging on anorectal and pelvic floor functions in females. *Dis Colon Rectum* 2006; 49: 1726–1735.
- Camilleri M, Iturrino J, Bharucha AE, et al. Performance characteristics of scintigraphic measurement of gastric emptying of solids in healthy participants. *Neurogastroenterol Motil* 2012; 24: 1076–e562.
- 17. Deiteren A, Camilleri M, Bharucha AE, et al. Performance characteristics of scintigraphic colon transit measurement in health and irritable bowel syndrome and relationship to bowel functions. *Neurogastroenterol Motil* 2010: 22: 415–423, e95.
- 18. Bouras EP, Delgado-Aros S, Camilleri M, et al. SPECT imaging of the stomach: comparison with barostat and effects of sex, age, body mass index, and fundoplication. *Gut* 2002; 51: 781–786.
- Kroenke K, Spitzer RL and Williams JBW. The PHQ-9: validity of a brief depression severity measure. *J Gen Intern Med* 2001; 16: 606–613.
- 20. Kroenke K and Spitzer RL. The PHQ-9: a new depression diagnostic and severity measure. *Psychiatric Annals* 2002; 32: 509–521.
- Löwe B, Unutzer J, Callahan CM, et al. Monitoring depression treatment outcomes with the Patient Health Questionnaire-9. *Med Care* 2004; 42: 1194–1201.
- 22. Spitzer RL, Kroenke K, Williams JBW, et al. A brief measure for assessing generalized anxiety disorder: the GAD-7. *Arch Intern Med* 2006; 166: 1092–1097.
- 23. Shin A, Camilleri M, Nadeau A, et al. Interpretation of overall colonic transit in defecation disorders in males and females. *Neurogastroenterol Motil* 2013; 25: 502–508.
- American Psychiatric Association. Diagnostic and Statistical Manual of Mental Disorders, 4th ed. Washington, DC: American Psychiatric Association, 2000.

- Nehra V, Bruce BK, Rath-Harvey DM, et al. Psychological disorders in patients with evacuation disorders and constipation in a tertiary practice. Am J Gastroenterol 2000; 95: 1755–1758.
- Lackner JM, Ma CX, Keefer L, et al. Type, rather than number, of mental and physical comorbidities increases the severity of symptoms in patients with irritable bowel syndrome. Clin Gastroenterol Hepatol 2013; 11: 1147–1157.
- 27. Khan S, Hyman PE, Cocjin J, et al. Rumination syndrome in adolescents. *J Pediatr* 2000; 136: 528–531.
- Green AD, Alioto A, Mousa H, et al. Severe pediatric rumination syndrome: successful interdisciplinary inpatient management. J Pediatr Gastroenterol Nutr 2011; 52: 414–418
- Bellini M, Rappelli L, Alduini P, et al. Pelvic floor dyssynergia and psychiatric disorders Does the snake bite its tail?. *Minerva Gastroenterol Dietol* 2003; 49: 135–139.
- 30. Rao SS, Seaton K, Miller MJ, et al. Psychological profiles and quality of life differ between patients with

- dyssynergia and those with slow transit constipation. *J Psychosom Res* 2007; 63: 441–449.
- 31. Beck JJ, Elzevier HW, Pelger RC, et al. Multiple pelvic floor complaints are correlated with sexual abuse history. *J Sex Med* 2009; 6: 193–198.
- Abraham S, Luscombe GM and Kellow JE. Pelvic floor dysfunction predicts abdominal bloating and distension in eating disorder patients. *Scand J Gastroenterol* 2012; 47: 625–631.
- Enck P, Van der Voort IR and Klosterhalfen S. Biofeedback therapy in fecal incontinence and constipation. Neurogastroenterol Motil 2009; 21: 1133–1141.
- 34. Gilliland R, Heymen S, Altomare DF, et al. Outcome and predictors of success of biofeedback for constipation. *Br J Surg* 1997; 84: 1123–1126.
- 35. Mason HJ, Serrano-Ikkos E and Kamm MA. Psychological state and quality of life in patients having behavioral treatment (biofeedback) for intractable constipation. *Am J Gastroenterol* 2002; 97: 3154–3159.