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REVIEW ARTICLE

Nonpharmacological treatment of rumination syndrome in childhood: A systematic review of the literature

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Abstract

Rumination syndrome (RS) is a complex functional disorder characterized by recurrent, repetitive regurgitation of recently swallowed food. RS may have medical and psychosocial implications, compromising the quality of life and causing high rates of school absenteeism. Pediatric RS has been poorly studied and little evidence regarding its treatment is available. This systematic review aims to evaluate the literature on the nonpharmacological treatment of RS in childhood. A systematic literature search was conducted on MEDLINE/PubMed, CINAHL, Cochrane Library, PsycINFO, and PEDro, from 2000 to 2023. The methodological quality of the publications was assessed by applying the guidelines proposed by the Equator network, according to the different designs of study, and the risk of bias was evaluated with the Risk Of Bias In Non-Randomized Studies of Interventions (ROBINS-I). Five hundred ninety-six studies were screened, and 7 studies were included in the review. Diaphragmatic breathing was the most used nonpharmacological treatment, and it was always combined with other therapeutic strategies. The vast heterogeneity of the physical or mental comorbidities and the methodology adopted in the publications did not allow a comparative analysis of the different treatments. Regardless of the type of treatment, high-intensity therapeutic programs and specific operators' training emerged as the most influencing factors for patients' outcomes. According to the available evidence, there is not enough high-quality evidence to suggest a defined therapeutic strategy. Large observational studies on selected patients accounting for possible confounders, with adequate follow-up times, and with clearly defined treatment regimens are needed to identify the best therapeutic approach.

KEYWORDS

behavioral treatment, diaphragmatic breathing, pediatric, rumination syndrome

1 | INTRODUCTION

Rumination syndrome (RS) is an acquired and unconscious behavior characterized by the recurrent, and repeated regurgitation of food soon after the meal that resolves within 1–2 h.[1–5] The simultaneous contraction of the abdominal and intercostal muscles induces the retrograde flow of gastric contents to the mouth; the food is then reswallowed or spitted.[1,6,7] The Rome IV criteria and the Diagnostic and Statistical Manual of Mental Disorders, 5th Edition (DSM-5) classify RS as both a "functional gastrointestinal disorder" and a "feeding and eating disorder"[8] (Tables S1 and S2).

RS can affect patients of all ages and cognitive abilities, with or without medical or psychiatric

comorbidities,[2,4,6] and its prevalence is estimated at up to 11.94% of children.[1,4,5,9–11]

RS is considered a disease with a favorable prognosis, but it may have medical and psychosocial implications, including secondary esophageal injury and school absenteeism,[3,12] thus compromising the quality of life. Therefore, rehabilitative interventions, including behavioral therapies, are often required.

Diaphragmatic breathing (DB) is one of the most used treatments for adults with RS.[10] DB is a controlled breathing technique, leading the patient to implement antagonistic behavior to the rumination. DB may be associated with biofeedback or electromyography of the abdominal and intercostal muscles to enhance its effects.[1,7,13,14] Chewing gum consumption and hypnosis have also been proposed.[1,14–16]

Few data are available for children. This systematic review aims to evaluate the available evidence on the efficacy of nonpharmacological treatments in the pediatric population affected by RS.

What Is Known

- Rumination syndrome (RS) is an often inaccurately diagnosed functional disorder characterized by a recurrent, repetitive regurgitation of recently swallowed food.
- RS can cause significant quality-of-life impairment.
- RS is treated with many nonpharmacological strategies, but there is not enough high-quality evidence to support one above the others.

What Is New

- Diaphragmatic breathing is the most used nonpharmacological treatment, always combined with other therapeutic strategies.
- High-intensity therapeutic programs and specific operators' training are crucial factors for patient's outcomes.
- Large observational studies with clearly defined treatment regimens on patients with homogeneous characteristics are needed.

2 | METHODS

This review follows the PRISMA standards on reporting on systematic review. The study did not require the ethics committee's approval because it consisted of examining the existing literature.

2.1 | Data sources and search strategy

A literature search was conducted on the following electronic database: MEDLINE/PubMed, CINAHL, Cochrane Library, PsycINFO, and Web of Science. The terms used in the search strategy are listed in Table S3.

2.2 | Study selection

The inclusion and exclusion criteria are reported in Table S4. Studies were identified for this systematic examination with a three-step process: 1- title screening, 2- abstract review, and 3- complete article review.

Studies including patients aged 5–21 years diagnosed with RS according to the criteria of the Rome Foundation or the DSM-5, published between January 1, 2000 and March 30, 2023, available in English and Italian were considered. Only study reporting a clear description of the treatment were included.

Case reports, case series with fewer than five patients, studies on adults including less than five patients aged <21 years, letters to the editor, systematic and narrative reviews, book chapters, and abstracts were excluded.

2.3 | Data extraction and quality assessment

The data of each eligible study were extracted, without making any modifications, and reported according to characteristics of the samples (size, comorbidities, associated symptoms), treatments (setting, duration, and intensity of treatment, the team involved), and possible confounding factors (follow-up times, outcomes, discontinuation criteria). Two authors (R. S. and A. D. T.) independently assessed the research reporting quality using the Equator Network guidelines Strengthening the reporting of Observational Studies in Epidemiology (STROBE), Case Reports Guidelines (CARE), and Consensus on Exercise Reporting Template (CERT). A third reviewer, A. T., resolved any disagreements.

In case of heterogeneity limiting the possibility of performing a direct comparison between the studies, an assessment of the study's methodological quality and the risk of bias was performed using the Risk Of Bias In Non-Randomized Studies-of Interventions (ROBINS-I).

3 | RESULTS

Seven hundred and ninety-one studies were found, with 11 additional publications taken from the studies' references. Duplicates were removed by reviewing and comparing abstracts. After the selection process,

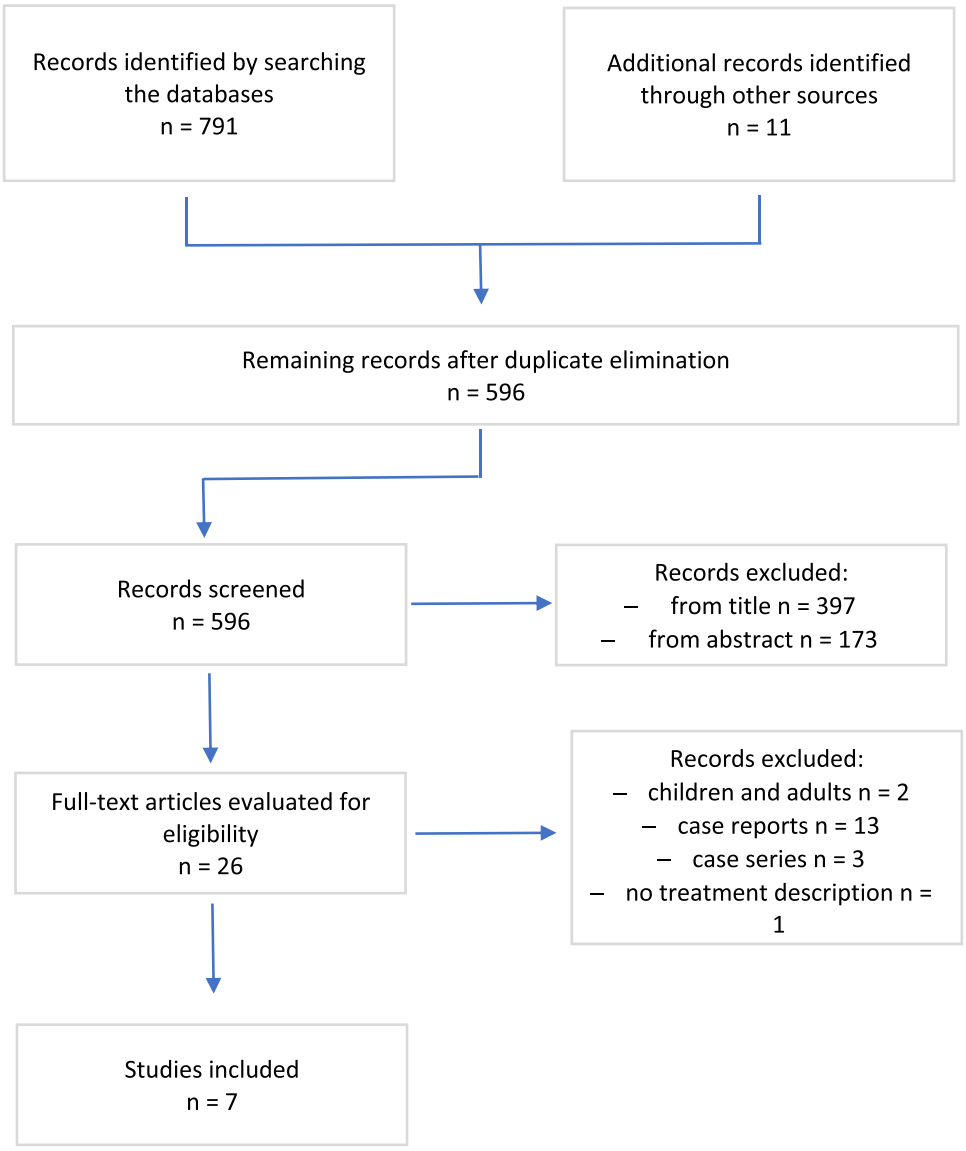

FIGURE 1 Study selection flowchart.

seven studies were included in the review (Figure 1): five studies retrospective,[3,12,17–19] and two prospective case series.[20,21] Quality assessment of the selected studies is shown in Table S5.

3.1 | Characteristics of the studies

The characteristics of the studies are reported in Table 1. The inclusion criteria adopted in the studies were very heterogeneous. Most of the patients had long-lasting and intense symptoms and medical comorbidities such as dehydration, weight loss, and organ function alterations that required complex care in hospital settings.[3,17,20] None of the studies considered medical and psychiatric comorbidities and disease severity as possible confounders.

3.2 | Characteristics of nonpharmacological treatment

Table 2 reports the treatment program proposed by each study. The 16-item CERT reporting exercise interventions is shown in Table S6.

DB was the most used treatment, even though it was combined with other therapeutic strategies (biofeedback,[3,17,20] cognitive-behavioral therapies,[3,17] relaxation training,[3,17] reswallowing regurgitation,[18–20] distraction strategies,[19,20] counseling,[3,12,17–19] slow pace of eating,[18,19,21] posture while eating,[18,19,21] smaller portions[19,21]).

In all studies except for one,[18] the treatments changed over time but the reason for the therapeutic switching was not specified. Despite the diversity of treatment choices, all the authors agreed to consider

TABLE 1 Characteristics of included studies.

Author year	Methodology	Sample number (female)	Sample characteristics	Psychiatric comorbidity	Medical comorbidity	Associated symptoms	Symptoms duration before diagnosis
Khan et al.[17]	Retrospective 1994–1998	12 (9F)	9–19 years Subjects without developmental delay or neurological lesions *N* = 2 do not accept the diagnosis	*N* = 3 (25%) depression *N* = 3 (25%) somatic symptom disorder *N* = 1 (8.33%) Asperger syndrome *N* = 1 (8.33%) chronic pain *N* = 2 (16.6%) suspected eating disorder	*N* = 1 (8.33%) neurofibromatosis (with depression) *N* = 8 enteral/parenteral feeding	*N* = 10 (83.3%) abdominal pain *N* = 7 (58.3%) weight loss >2 kg *N* = 4 (33.33%) nausea *N* = 2 (16.6%) abdominal bloating	1–72 months
Chial et al.[3]	Retrospective 1975–2000	147 (68% F)	5–20 years 46% hospitalized In 127 of 147 suggested treatment 71 treated by the research institute 54 (36.7%) are known for their outcome None requires tube feeds *N* = 2 received only counseling	*N* = 3.4% eating disorder *N* = 5 (3.40%) intellectual disability *N* = 2 (1.36%) developmental disabilities *N* = 1 (0.68%) autism *N* = 1 (0.68%) dyslexia	*N* = 1 Bartter's syndrome	*N* = 42.2% weight loss (median 7 kg) *N* = 38% abdominal pain *N* = 21% constipation *N* = 17% nausea *N* = 8% diarrhea	2.2 ± 0.3 years
Green et al.[20]	Prospective case series	5 (4F)	14–20 years *N* = 4 enteral/parenteral feeding None with psychiatric comorbidity or eating disorders	/	/	*N* = 5 weight loss (range 7–23 kg)	5–48 months
Malik et al.[12]	Prospective 01/2012 to 12/2013	50 30 with RS (11F)	12.4 ± 2.4 mean age	*N* = 5 (16.6%) anxiety, mild intellectual disability, learning disabilities, facial tic *N* = 2 (6.6%) eating disorder	*N* = 2 gastroesophageal reflux *N* = 1 malrotation *N* = 1 functional dyspepsia	*N* = 9 (30%) nausea *N* = 7 (23.3%) constipation *N* = 7 (23.3%) abdominal pain	2–72 months

TABLE 1 (Continued)

Author year	Methodology	Sample number (female)	Sample characteristics	Psychiatric comorbidity	Medical comorbidity	Associated symptoms	Symptoms duration before diagnosis
Hawa et al.[18]	Retrospective 2018–2020	34 (79% F) 33	7–19 years	*N* = 17 (50%) anxiety disorder *N* = 7 (21%) depression *N* = 5 (15%) eating disorder *N* = 5 (15%) functional abdominal pain	*N* = 6 (18%) gastroesophageal reflux *N* = 4 (12%) superior mesenteric artery syndrome *N* = 9 (26%) need for feeding tube or parenteral nutrition	/	/
Lamparyk et al.[21]	Retrospective case series 12/2015 to 03/2020	28 23 included for criteria selection (47.8% F)	8–18 years Excluded autistic subjects Excluded subjects with eating disorders	*N* = 4 anxiety disorder *N* = 1 depression *N* = 1 bipolar disorder *N* = 1 obsessive compulsive disorder	*N* = 4 gastroesophageal reflux *N* = 3 pain associated disability syndrome (PADS) *N* = 2 asthma *N* = 1 irritable bowel syndrome *N* = 1 type 1 diabetes	/	/
Sabella et al.[19]	Retrospective 2018–2020	197 171 included for criteria selection (64%F)	9–17 years Exclusion criteria: intellectual disability; autism; infantile cerebral palsy 21% of the patients and/or family do not accept the diagnosis	Anxiety disorder: outpatient program (OP) 32%; intensive outpatient program (IOP) 50%; intensive inpatient program (IP) 36% Depression: OP14% IOP 14%; IP 20% Eating disorder: OP 5%; IOP 5%; IP 8%	Gastroesophageal reflux: OP20%; IOP 23%; IP 28% Gastroparesis: OP 1%; IOP 5%; IP 20% Irritable bowel syndrome: OP 6%; IOP 5%; IP 16% Feeding tube: IOP 18%, IP 56%	/	2–3 years

Abbreviations: F, female; RS, rumination syndrome (recurrent, repetitive regurgitation of recently swallowed food).

TABLE 2 Proposed treatment.

Author years	Setting	Treatment	Duration/intensity of treatment	Equipe	Defined stop therapy	Follow-up
Khan et al.[17]	Hospital	1. Behavioral Rehabilitation • Psychotherapy • Diaphragmatic breathing/biofeedback • Relaxation techniques 2. Pain management (if necessary)	5–36 months	Gastroenterologist Psychologist Dietitian Pain-management specialist	/	Stop therapy 5–36 months
Chial et al.[3]	Outpatient clinic 35 of 76 hospitalized (46.1%)	1. Behavioral treatment: • Biofeedback • Relaxation training • Diaphragmatic breathing 2. Cognitive-behavioral treatment 3. Counseling	10.2 ± 1.4 months Intensity: 1–19 sessions	/	/	Stop therapy 10.2 ± 1.4 months *N* = 1 lost/dead (Bartter's syndrome)
Green et al.[20]	Hospital	Psychology protocol: 1. Biofeedback (*N* = 1) 2. Reswallowing food 3. Distraction strategies (conversation, walking, playing, video games, reading; *N* = 3) 4. Diaphragmatic breathing (*N* = 5)	9–31 gg Intensity: three sessions each day; from 30 min to 2 h duration	Gastroenterologist Psychologist Dietitian Child life specialist Therapeutic recreation Physiotherapist	Resuming oral feeding	Stop therapy 9–31 days
Malik et al.[12]	Outpatient clinic	*N* = 23 (77%) 1. Diaphragmatic breathing plus behavioral therapy 2. Counseling (*N* = 7, 23%)	Mean duration 30 days (range 2–90 days)	/	/	Stop therapy 14.7 ± 4.6 months
Hawa et al.[18]	Outpatient clinic telemedicine	Intensive medical and behavioral treatment: 1. Direct observation of food intake (with real time feedback) 2. Behavioral strategies: • Diaphragmatic breathing • Reswallowing regurgitation • Slow pace of eating • Posture while eating	Mean duration 4 days Intensity: 2–3 sessions each day; 60 min per day	Gastroenterologist Psychologist Dietitian	/	First follow-up: stop therapy Second follow-up: in July 2021 (time interval not known) Lost *N* = 3 followed by telemedicine, while *N* = 7 followed in person
Lamparyk et al.[21]	Outpatient clinic	Comprehensive behavioral treatment for rumination syndrome (CBT-R): 1. Teaching skills (to control the rumination itself) • Diaphragmatic breathing • Posturing 2. Rumination elimination: • Reducing volume of oral intake 3. Return to baseline: • Develop specific plan to gradually progress oral intake • Supplemental nutrition if necessary	From <1 to 69 weeks Intensity: 1–22 sessions	Psychologist	/	Stop therapy 1–69 weeks

TABLE 2 (Continued)

Author years	Setting	Treatment	Duration/intensity of treatment	Equipe	Defined stop therapy	Follow-up
		4. Phase out-interventions and relapse prevention • Reduce reliance on diaphragmatic breathing by increasing awareness of premonitory urge and target competing responses				
Sabella et al.[19]	Outpatients clinic hospital	Sample is divided in three subpopulations, with different approaches, considering: • Domicile (near or far from the reference center) • Symptoms severity • Treatment refractory 1. OP: near the center, medium severity of symptoms 2. IOP: far from the center, greater symptoms severity and/or OP treatment refractory 3. IP: severe symptoms and/or IOP treatment refractory Outpatient program (OP) 1. Psychotherapy (52%) 2. Behavioral treatment: Diaphragmatic breathing Reswallowing regurgitation Smaller portion Pacing meals Distraction after meals Intensive Outpatient (IOP) and Inpatient program (IP) 1. Psychotherapy 2. Behavioral treatment: Diaphragmatic breathing Reswallowing regurgitation Smaller portion Pacing meals Distraction after meals	OP One session every 2 or 4 weeks Duration: 45–60 min IOP/IP Three session everyday Duration: 45–60 min	Gastroenterologist Psychologist Dietitian Child life specialist Therapeutic recreation Physiotherapist	Symptoms improvement/ resolution	First follow-up: stop therapy Lost $N = 48$ Second follow-up: 5.3 months after stop therapy (median) $N = 16$ (IOP 7, IP 9)

Abbreviations: IP, intensive inpatient program; IOP, intensive outpatient program; OP, outpatient program.

therapeutic education and empowerment strategies as two fundamental phases of the treatment process.[3,12,17–21] Four studies involved multiprofessional teams,[17–20] including psychologists,[17–20] gastroenterologists,[17–20] dieticians,[17–20] pain therapy specialists,[17] physiotherapists,[18–20] child life specialist,[18–20] recreational therapists.[18–20] The remaining articles did not provide information.

The duration and intensity of the treatments were highly variable, even within the same study (treatments durations ranging from 4 days to 69 weeks[18] and intensity from 2 h three times a Day 3 to 1 h every 2–4 weeks[19]). Despite this, treatment intensity was often reported as crucial in determining the outcome.[3,19,21]

Only one study reported the reasons for treatment suspension.[20] The last follow-up, which ranged from 1 week to 36 months,[17,21] coincided with the end of the therapeutic intervention in five studies,[3,12,17,20,21] while considered a second evaluation after a mean of 5.3 a 12 months was performed in two studies.[18,19]

3.3 | Outcomes of nonpharmacological treatments (Table 3)

The included studies reported the outcomes expressed as "resolution,"[3,12,17–21] "improvement,"[3,12,17–21] "positive impact on symptoms,"[3] "partially responding,"[12] "nonresponders,"[3,12,17–21] "worsened,"[18,19] "relapsed"[12,17,21] on different variables rumination,[17] reswallowing,[18,19] vomiting,[18,19] number of not-consumed meals,[17–20] need for enteral or parenteral food supplementation,[18–20] weight loss,[18–20] and loss of social and school activities.[17,19] The findings were heterogeneous: remission ranged from 0%[18] to 100%,[17] improvement from 8.33%[17] to 87.5%,[18] and nonresponse from 4.2%[18] to 16.6%.[17]

3.4 | Risk of bias

The effectiveness of interventions was estimated by evaluating the risk of bias through the ROBINS-I tool (Table S7).

Only two articles described the statistical methods used for collecting and interpreting the data,[12,19] and none considered the external validity of the data. Two failed to report the outcomes completely (reporting bias).

One study involving a large retrospective cohort had an observation period of 25 years, and given the long follow-up period, the findings could be affected by a recall bias.[3]

In three studies,[3,18,19] patients were still on treatment at the end of the follow-up (attrition bias), while in two[3,19] 40% or less of the selected patients were included in the analysis due to data loss.

4 | DISCUSSION

This study was the first to systematically review the nonpharmacological treatments for RS in childhood.

DB emerged as the most commonly used treatment and could be considered a harmless general approach. DB is an easy-to-learn technique that is well-known to provide effective and well-tolerated therapy for adult patients with RS, as well documented with high-resolution manometry after consumption of a meal.[22] DB operates as a competing response to habitual abdominal wall contraction by relaxing the abdominal wall.[23] However, DB requires experience to be taught and children and adolescents must be cooperative and have sufficient cognitive tools to learn the technique. Therefore, an inadequate performance of DB could explain its ineffectiveness in certain cases.

Interestingly, this review also showed that DB was generally offered combined with other nonpharmacological strategies mainly referring to cognitive-behavioral or relaxation treatments.

The use of these strategies suggests that all the authors recognize a behavioral problem underlying the disorder and the treatment should focus not only on the rumination per se but on the complete mental and physical well-being of the patient.

Indeed, RS is a complex functional disorder, with most of the patients affected with RS presenting chronic and intense symptoms, associated with other prognostic factors (medical and psychiatric comorbidities,[12,17,19,20] weight loss,[3] acceptance of the diagnosis[17,19]). The severity of symptoms and comorbidities may affect the setting of treatment,[3,12,17–21] suggesting the need for an inpatient multiprofessional management for complex cases.

Conversely, less severe patients can be managed with self-performed treatments at home. However, the latter approach lacks of a strict supervision which may influence the quality of the intervention. In the included studies, no data about compliance were available, but it is plausible that some patients did not perform the assigned exercises continuously.

Thereby, it can be supposed that it is necessary to carefully calibrate the treatment modalities according to the specific clinical scenario to optimize the efficacy of the interventions.

Overall, treatment intensity was crucial in determining the outcome.[3,19,21] Unfortunately, the wide span between the treatment duration within the studies did not allow for identifying indications of its optimal values. Anyhow, available data suggested that a high-intensity program (three sessions/day) in the short term could be more effective than a low-intensity one (one to two sessions/week) over a long time.

This systematic review has some limitations, starting from the small number of the studies and patients included.

TABLE 3 Summary results of included studies.

Author year	Outcome	Rumination syndrome severity outcomes	Results				
			Resolution	**Improvement**	**Nonresponder**	**Worsening**	**Recurrence**
Khan et al.[17]	Decrease in episodes frequency Rumination Resuming oral feeding Return to routine activities/school	/	*N* = 9 (75%) Regurgitation *N* = 8 (100%) Oral feeding *N* = 12 (100%) Routine activities/ school	*N* = 1 (8.33%)	*N* = 2 (16.6%) (1 with fibromatosis; 1 not accepting diagnosis)	/	*N* = 1 Sporadic
Chial et al.[3]	/	/	*N* = 16 (29.6%)	*N* = 30 (55.5%)	*N* = 12.9%	/	/
Green et al.[20]	Resuming oral feeding	1. Weight loss 2. Requiring feeding tube	*N* = 5 (100%)	/	/	/	/
Malik et al.[12]	/	/	*N* = 19 (82.6%)	*N* = 2 (8.7%) Partial response (?)	*N* = 2 (8.7%)	/	*N* = 8 (26.6%)
Hawa et al.[18]	1. Daily vomiting 2. Daily reswallowing 3. Skipping meals 4. Requiring feeding tube 5. Losing weight	Reswallowing/vomiting were scored on a Likert scale from 1 to 6: 1. Everyday 2. Several times a week 3. Once a week 4. Once every 2–3 weeks 5. Once a month 6. Never Frequency: 1. 0–5 times per day 2. 5–10 times per day 3. 10 or more times per day Skipped meals: <1 time/day 1 time/day 2 times/day 3–4 times/day Need for feeding tube: yes or no	First follow-up In person: *N* = 16.7% Telemedicine: *N* = 0% Symptom severity Daily vomiting Daily reswallowing Skipping meal Requiring feeding tube Losing weight	First follow-up In person: *N* = 79.2% Telemedicine *N* = 87.5% First follow-up *N* = 33 6.1% 30.3% 0% 6.3% 0%	First follow-up In person: *N* = 4.2% Telemedicine: *N* = 12.5% Second follow-up *N* = 23 17% 52% 39% 9% 13%	Second follow-up *N* = 23 (100%)	/
Lamparyk et al.[21]	/	Frequency	*N* = 73.9%	*N* = 13%	*N* = 3	/	*N* = 17.4%

(Continues)

TABLE 3 (Continued)

Author year	Outcome	Rumination syndrome severity outcomes	Results				
			Resolution	Improvement	Nonresponder	Worsening	Recurrence
Sabella et al.[19]	1. Daily vomiting 2. Daily reswallowing 3. Skipping meals 4. Requiring feeding tube 5. Losing weight 6. Attending school 7. Attending social events	Frequency of 1, 2, and 3	Follow-up stop therapy OP $N = 14\%$ IOP $N = 21\%$ IP $N = 9\%$ Follow-up after 5.3 months IOP $N = 0\%$ IP $N = 27\%$	Follow-up stop therapy OP $N = 57\%$ IOP N = 74% IP $N = 87\%$ Follow-up after 5.3 months IOP $N = 86\%$ IP $N = 45\%$	Follow-up stop therapy OP $N = 26\%$ IOP $N = 5\%$ IP $N = 4\%$ Follow-up after 5.3 months IOP $N = 0\%$ IP $N = 11\%$	Follow-up stop therapy OP $N = 3\%$ IOP $N = 0\%$ IP $N = 0\%$ Follow-up after 5.3 months IOP $N = 14\%$ IP $N = 22\%$	/

Moreover, due to the inclusion criteria, some treatment modalities, such as chewing gum consumption or hypnosis, which have some evidence of efficacy both in children and adolescents[1,14–16] were not examined.

Furthermore, the methodological heterogeneity of the included studies did not allow us to analyze the treatment efficacy according to the physical or mental comorbidities and to assess the superiority of one treatment strategy over the others.

Despite this limitation, this review offers a wide view of the most used nonpharmacological strategies for treating RS in children.

5 | CONCLUSION

Intensive programs of DB combined with other cognitive behavior or relaxing techniques may be efficient for treating children with RS but there is no high-quality evidence to suggest a defined therapeutic strategy.

Larger observational studies on selected patients accounting for possible confounders (disease severity or presence of medical-psychiatric comorbidities) and with adequate follow-up times are needed. Moreover, specific tools are needed to measure and monitor treatment adherence, using outcome assessment tools capable of measuring activity and participation levels according to the model of the International Classification of Functioning, Disability and Health for Children and Youth.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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