

preliminary findings are a promising indicator that DCS is an effective adjunct to behavioural intervention, although larger clinical trials are warranted to fully verify this.

3.2 Psychological treatment

Five case studies were found to report on the use of cognitive behavioural therapy (CBT) to treat ARFID. In four studies, the interventions used CBT approaches to formulate and address eating-associated anxiety and fears about food consumption, without the focus on weight and shape concerns used in CBT methods for other eating disorders such as AN [13-16]. A fifth study employed a novel 4-week, exposure-based CBT intervention, developed to target other drivers of food avoidance and/or restriction (i.e., disgust sensitivity, dysfunctional cognitions about feared foods, the aversive consequences of eating) [17]. This method, which has been designed specifically for adolescents with ARFID and integrates inhibitory learning principles has demonstrated preliminary success in treating a number of ARFID presentations.

Cognitive Behavioural Therapy (CBT): is a short-term, goal-oriented psychotherapy treatment that takes a hands-on, practical approach to problem-solving. Its goal is to change patterns of thinking or behaviour that are behind people's difficulties, and so change the way they feel. CBT works by changing people's attitudes and their behaviour by focusing on the thoughts, images, beliefs and attitudes that are held (a person's *cognitive processes*) and how these processes relate to the way a person behaves, as a way of dealing with emotional problems

Examples: Learning how to manage stress and anxiety (e.g., learning relaxation techniques such as deep breathing, coping self-talk such as "I've done this before, just take deep breaths," and distraction) identifying situations that are often avoided and gradually approaching feared situations.

Two case series and one feasibility study were found to report on the use of family-based therapy (FBT) to treat ARFID [18-20]. FBT, which is designed to empower caregivers, reduce familial guilt and support recovery at home, is often used in the treatment of eating disorders. Although FBT-ARFID is similar in this respect, and employs the main principles of FBT, it has been adapted to address the needs of patients with different ARFID presentations, targeting those with sensory sensitivities, fear-based concerns and little interest in eating [18]. Though limited by small sample sizes and lack of a long-term follow up, the evidence suggests that FBT may prove to be a feasible treatment approach. In a

similar manner, a small number of parent training curricula have been trialled which aim to coach caregivers in implementing at-home behavioural feeding interventions. Initial findings indicate that both parent teleconsultation and attendance at group education sessions can adequately prepare caregivers to support children who engage in severe selective eating but do not require treatment in a hospital setting [21, 22].

Family-based therapy (FBT) for eating disorders is commonly known as The Maudsley Model and was originally developed to treat adolescents with Anorexia Nervosa and Bulimia Nervosa. FBT aims to assist the family, namely the parents, to bring about recovery in their child with an eating disorder. The core principles of are:

1. No one is to blame for the development of the eating disorder
2. The eating disorder is externalised or separated from the sufferer and the eating disorder is targeted to reduce blame and criticism
3. The family are viewed as the best resource to bring about recovery
4. Hospitalisation is a short term solution for the problem
5. Each family member is assigned a specific role

3.3 Multi-modal approach

Intervention-focused papers commonly endorse a multi-modal approach, characterised by input from a multidisciplinary team and incorporating a wide range of interventions [23, 24, 20]. The efficacy of such an approach was supported by an RCT investigating the treatment of chronic food refusal in a day treatment programme [25]. The researchers randomly assigned twenty children aged 13–72 months to either a waiting list or a **five-day intensive behavioural intervention** with treatment input from a multidisciplinary team. *Despite a small sample, the intervention group displayed significantly greater improvements ($p < .05$) on all primary outcomes compared to no treatment, suggesting that a collaborative approach to treatment can safely and effectively address the challenging nature of food refusal.*

Table 1. Summary of ARFID articles relating to treatment

Author (year) and country	Study aim	Methodology and sample	Symptoms/presentation	Treatment	Outcome
<u>Pharmacological treatment</u>					
Brewerton & D'Agostino 2017 [8] USA	To document the clinical progress of ARFID patients treated with low doses of adjunctive olanzapine	Retrospective chart review of 9 patients (8 females and 1 male) (9–19 years) - Mean admission BMI 15.6 ± 1.8 kg/m ²	Participants diagnosed with ARFID using DSM-5 criteria	- Adjunctive low-dose olanzapine (alongside meal behaviour therapy and other treatment modalities offered to ED patients) - Mean number of days on olanzapine 53.4 ± 22.4	- Mean change in BMI 3.1 ± 1.34 kg/m ² - Mean change in BMI index for-age percentile 11.0 ± 14.7 to 35.9 ± 27.5 Olanzapine promoted weight gain in all patients and relieved symptoms of anxiety, depression and cognitive impairment
Okereke 2018 [9] USA	To describe the successful treatment of anxiety using Buspirone in an individual with ARFID	Case study 14-year-old female BMI 20.3 kg/m ² (58 th percentile)	Complaints of anxiety, abdominal pain and vomiting resulting in food restriction (later diagnosed with ARFID as well as irritable bowel syndrome)	- Individual and family therapy - Sertraline at 50 mg/day (discontinued when patient experienced agitation and thoughts of suicide) Buspirone 5 mg twice daily increased to 7.5 mg twice daily at 1 month follow up and 10 mg twice daily at 6-month follow-up - Follow-up 1, 2, 4, 6, and 8- months post-treatment	- BMI at 8-month follow up was 22.0 kg/m ² (73 rd percentile) - SSRIs can be used to treat eating-related anxiety but may cause adverse side effects, particularly in children and adolescents - Buspirone successfully treated anxiety symptoms associated with eating (patient denied any significant side effects)

<p>Tanidir & Herguner 2015 [10]</p> <p>Turkey</p>	<p>To present a case of ARFID successfully treated with mirtazapine</p>	<p>Case study 10-year-old female</p> <p>Weight 26 kg on admission (below 10th percentile)</p>	<p>Refusal to eat solid food after choking incident at 4 years old</p>	<ul style="list-style-type: none"> - Initial behavioural approach - 10 mg/day fluoxetine increased over time to 30 mg/day for 2 months with no success - 15 mg/day mirtazapine for 6 months 	<ul style="list-style-type: none"> - Weight increased to 34 kg (25–50th percentile) - Mirtazapine well tolerated - marked and rapid improvement in symptoms relating to choking phobia - Within 2 weeks, the patient reported less anxiety during mealtimes and experienced an increase in appetite - No re-emergence of complaints at 6-month follow up
<p>Gray 2018 [11]</p> <p>USA</p>	<p>To evaluate the use of mirtazapine in treating patients with ARFID</p>	<p>Retrospective chart review 6 females, 8 males (7–23 years) who received treatment at a San Diego eating Disorders clinic from 2015 to 2016. Mean BMI at intake 16.8 ± kg/m²</p>	<p>Difficulty eating related to low appetite cues, taste, or texture sensitivity, anxiety of an adverse event (e.g., choking), or significant functional gastrointestinal distress</p>	<ul style="list-style-type: none"> - Six patients treated with mirtazapine as monotherapy and 8 on additional medications - Average dose of mirtazapine 25.5 mg - Follow-up 6-months post treatment and monthly follow ups thereafter 	<ul style="list-style-type: none"> - Average change in BMI without mirtazapine = 0.10 BMI point per week - Average change in BMI with mirtazapine = 0.23 BMI point per week ($t_{13} = -3.11, p < .05$) - Overall, mirtazapine was safe, well tolerated and encouraged greater weight gain than treatment-as-usual programme
<p>Sharp 2017 [12]</p> <p>USA</p>	<p>To examine the feasibility and preliminary efficacy of combining D-cycloserine with a behavioural intervention in treating young children with chronic food refusal</p>	<p>Double-blind, placebo controlled study 16 children (37.5% female) 18 months – 6 years</p>	<p>Active and persistent food refusal which severely restricted the volume of food consumed</p>	<ul style="list-style-type: none"> - Randomisation to intensive Behavioural intervention + D-cycloserine OR intensive behavioural intervention + placebo over 5 days (15 meals in total) - Follow-up 1-month post treatment 	<p>Mealtime behaviours improved significantly in both groups, but D-cycloserine further enhanced response to intervention, rapidly increased food acceptance and reduced disruptive behaviours</p>

<u>Psychological treatment</u>					
Fischer 2015 [13] USA	To evaluate the effects of an intervention for chronic food selectivity in an adolescent with ARFID	Case study 16-year-old-male	History of extreme food selectivity, associated feeding anxiety and some acute sensory aversion to certain foods	<ul style="list-style-type: none"> - Intervention incorporating both a clinic (behavioural treatment and CBT) and concurrent in-home component (enforced by the patient's mother) - Follow-up 1- and 3-month post treatment 	<ul style="list-style-type: none"> - Greater consumption of foods (both quantity and variety) - Reduced anxiety and ability to eat out in a social environment - Daily bowel movements and increased energy (findings maintained post-treatment)
King 2015 [14] USA	To present a case of ARFID successfully treated with CBT	Case study 41-year-old female, BMI 15.5 kg/m ²	Patient had Crohn's disease as a child and developed severe illness anxiety following acute gastroenteritis which caused her to limit food intake	<ul style="list-style-type: none"> - Inpatient treatment - 8 sessions of CBT including psychoeducation, systematic desensitisation (in vivo exposure) and cognitive restructuring - Follow-up 8-months post treatment 	<ul style="list-style-type: none"> - At discharge, patient was consuming 1650 calories daily and BMI 16.5 kg/m², and reported reduced anxiety and increased energy - At 8 months post-discharge, patient BMI was 19.4 kg/m²
Aloi 2015 [15] Italy	To present a case of ARFID successfully treated with CBT and family involvement	Case study 24-year-old male, slightly overweight with BMI 25.5 kg/m ²	<ul style="list-style-type: none"> - Dysfunctional eating behaviours dating back to the age of 2 - Avoidance based on an unpleasant sensory experience - Complaints of anxiety relating to shared meals, resulting in social withdrawal 	<ul style="list-style-type: none"> - Psychotherapeutic intervention once a week for one hour over six months - Phase 1 (session 1–4) psychoeducation - Phase 2 (session 5–7) family Therapy - Phase 3 (session 8–18) CBT - Phase 4 (session 19–20) relapse prevention Follow up 6 months post treatment 	<ul style="list-style-type: none"> - Many new foods introduced to the patient's diet - Improved social relationships and willingness to engage in shared meals
Gormez 2018 [16] Turkey	To present a case of ARFID successfully treated with CBT	Case study 27-year-old female BMI 16 kg/m ² (lost 6 kg	Nausea, retching, vomiting and unable to tolerate the sight and smell of food	<ul style="list-style-type: none"> - 12 40-minute weekly CBT sessions as an inpatient and 8 sessions as an outpatient as 	4kg gained (bmi 17.5 kg/m ²). a further 2 kg gained (bmi 18.3 kg/m ²) 6-



		in the past 2 months		well as psychoeducation and dietary supervision - Also 30–45 mg of mirtazapine	months post discharge • Improvement on cognitive domains, energy levels and anxiety
Dumont 2019 [17] <i>Netherlands</i>	To test a new 4-week exposure-based CBT day treatment for adolescents with ARFID	Case series Patients referred to SeysCentra, a Specialised treatment facility for children with feeding disorders (<i>n</i> = 11), 36% female, 10–18 years	Various presentations including anxiety driven (phobia), lack of interest in food, driven by disgust or aversion	- Exposure based CBT treatment designed to address a variety of ARFID presentations (i.e., disgust sensitivity, distorted cognitions about the consequences of eating feared foods) - A non-concurrent multiple baseline design followed by 4-week CBT - Various measures taken at baseline and throughout including measurement of DSM-5 ARFID diagnosis, food neophobia, body weight and anxiety - Follow-up 3-months post treatment	-At follow up, 10 of the 11 patients were at a healthy weight and had an age adequate nutritional intake - For most, food neophobia scores decreased to a nonclinical range - Dysfunctional cognitions about food intake/eating and anxiety decreased - Tube feeding eliminated in 6 Patients - All 11 patients demonstrated a more varied food repertoire - Demonstrates a CBT approach which has the potential to treat various issues which drive restrictive/avoidant eating behaviours in ARFID
Lock 2018 [18] <i>USA</i>	To illustrate the use of FBT in treating preadolescents with ARFID	Case study (1) 8-year-old female (2) 9-year-old female (3) 11-year-old female	3 different ARFID presentations: (1) Low appetite and lack of interest in eating (2) Sensory aversion to food (3) Fear of eating and extreme fear of vomiting	Family Based Therapy	(1) No major changes in interest in food but capable of eating sufficient quantities and eating-related family conflicts decreased (2) Greatly increased range of food, increased flexibility in social situations (3) Coping strategies used to manage fears, steady weight

					gain and increased participation in school and social activities
Lock 2019 [19] USA	To assess the feasibility of conducting an RCT comparing FBT-ARFID to usual care Usual care = whatever medical or psychological treatments they chose for a period of 3 months exclusive of FBT	Feasibility study 28 children (5–12 years) and their families	Patients meeting DSM-5 criteria for diagnosis of ARFID	- Participants randomised to receive immediate treatment with FBT for ARFID or usual care for a period of 3 months (and then offered FBT-ARFID) -Dose and duration of treatment were allowed to fluctuate according to clinical need	- Effect size differences on measures of weight and clinical severity of symptoms were moderate to large, favouring FBT-ARFID over usual care - Improvements also observed in parental self-efficacy - An RCT comparing FBT-ARFID and usual care would be feasible
Bloomefield 2019 [21] USA	To examine the use of teleconsultation in treating a patient with ARFID	Case study 8-year-old-male	Frequent refusal of non-preferred foods resulting in tantrum behaviour (whining, crying, gagging) upon sight or smell	- Parent teleconsultation (behavioural feeding intervention to increase food variety) - Follow-up 1- and 4-months post-treatment	Increase in the frequency of bites of non-preferred foods
Dahlsgaard & Bodie 2019 [22] USA	To report the acceptability, feasibility and initial outcomes of the Picky Eaters Clinic	Pilot trial 21 children with a diagnosis of ARFID (4–11 years) and their Parents	Picky eaters (eating less than 20 foods, difficulty socialising, refusal to eat non-preferred foods)	-7 sessions (90 min each) of parent-led behavioural intervention - Follow-up 3-months post treatment	Reduction in picky eating and negative mealtime behaviours
Zucker 2018 [23] USA	To present an acceptance-based interoceptive exposure treatment for young people with ARFID and demonstrate its success in treating a	Case study 4-year-old female	- Patient had percutaneous endoscopic gastrostomy (PEG tube) since 14 months of age - Indifference to food, lack of awareness of	8 weekly sessions followed by 4 bimonthly sessions of acceptance based interoceptive exposure treatment - Feeling and Body Investigators (FBI)-ARFID Division (also mirtazapine for a month prior to exposure treatment)	- Patient no longer met criteria for ARFID - Notable improvement in capacity to cope with change, unknown internal sensations no longer viewed as a threat - Increase in quantity of food

	young girl with lifelong poor appetite		hunger, difficulty adjusting to a change in routine		consumed and need for supplemental feeds reduced - PEG tube eventually removed
<u>Multi-modal approach</u>					
Murphy & Zlomke 2016 [24] USA	To describe a behavioural feeding intervention used to treat a patient with ARFID	Case study 6-year-old female BMI 81st percentile (normal range)	- Gastroesophageal reflux disease - Began food refusal at 9 months old - Selective about food based on type, colour, texture, flavour and brand	- Behavioural feeding intervention with parent training strategies - Follow-up 6-weeks post treatment	Increased dietary repertoire and clinically significant decrease in problematic child and parent feeding behaviours
Lenz 2018 [25] USA	To describe the successful use of an intensive inpatient behavioural intervention in treating ARFID	Case study 8-year-old female diagnosed with ARFID	- Initially presenting with abdominal pain, nausea and vomiting which caused acute food refusal - Patient also stopped drinking fluids following a choking incident, which resulted in the placement of a nasogastric tube	- Initial outpatient treatment which employed family and individual therapy within a CBT framework - Subsequent inpatient admission to adolescent medicine service 16 outpatient sessions over a 12-week period and a 6-day inpatient stay - Follow-up 4-months post discharge	- Patient weight increased from lowest 21.8 kg to 26.5 kg (52nd percentile) at 4-month follow up - Full remission of ARFID symptoms
Spettigue 2018 [20] Canada	To examine the efficacy of treating ARFID patients with modified FBT or psychopharmacological treatment	Case series 5 females and 1 male (10–14 years)	Various presentations including fear following choking incident, abdominal pain and nausea, problems concentrating and severe anxiety	- Family Based Therapy - Medication –Olanzapine, Fluoxetine and Cyproheptadine - CBT	All six patients achieved their goal weight
Sharp 2016 [26]	To investigate the feasibility and	RCT at a	Children exhibiting active and persistent food refusal with	- Manual based and technology supported behavioural feeding intervention	- Children assigned to iEAT showed significantly greater improvements on

USA	preliminary efficacy of an intensive, manual-based behavioural feeding intervention for patients with chronic food refusal and/or dependence on enteral feeding	multidisciplinary day treatment programme ($n = 20$), 40% female, 13–72 months	dependence on enteral or oral supplementation	<ul style="list-style-type: none"> - integrated eating aversion treatment (iEAT) - iEAT vs. waiting list control (10 children randomised to each condition) - 14 40-minute meal blocks across 5 consecutive days (meals 1–11 with trained therapists and 12, 13 and 14 parent-led) - Follow-up 1-month post treatment 	<p>all primary outcome measures compared with controls</p> <ul style="list-style-type: none"> - At post-treatment follow up, all caregivers reported high levels of overall satisfaction with treatment
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One further study which investigated FBT for the treatment of ARFID has been published since the systematic review by Bourne et al (2020) [5] (Table 2).

Table 2: Additional published ARFID treatments

Author (year) and country	Study aim	Methodology and sample	Symptoms/presentation	Treatment	Outcome
Rienecke et al. (2020) [27] USA	To describe three different presentations of ARFID and how each responded to a family-based partial hospitalization program (PHP) for eating disorders	Case series 3 children with ARFID	<p>#1: ARFID following 2x choking incidents</p> <p>#2: extreme sensitivity to the taste and texture of food and significant anxiety around trying new foods. Reflux, vomiting, and colic, as well as pica at the age of 2 years</p>	<p>PHP based on Family Based Therapy (FBT) principles. Assigned a paediatric feeding psychologist who uses ABA and behavioural parent training.</p> <p>#1: Prompted by staff and parents to take small bites when noticing she was struggling to swallow. -Taught relaxation strategies such as deep breathing -22 treatment days</p> <p>#2: Positive and negative reinforcement. Small exposure to new foods. Response cost and negative punishment</p>	<p>All patients gained weight. No other objective or quantitative measure of improvements.</p> <p>An approach with emphasis on parental involvement seems promising, although research is needed to investigate this more fully</p>



			<p>#3: general disinterest in food and eating, as well as limited variety. Anxiety and depression</p>	<p>- 19 treatment days</p> <p>#3: Psychologist encouraged mother to increase food variety, calories, and consistency in her interactions during meal times</p> <p>-19 days in PHP</p> <p>- 12 days in Intensive outpatient program</p>	
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3.5 Discussion points

There are no well-established treatments for ARFID, with a limited number of randomized clinical trials among patients with ARFID. This literature review evidences several promising treatment avenues which warrant further study:

- 3) FBT, CBT and adjunctive pharmacological intervention appear to be the methods with the best evidence.
- 4) A multi-modal approach is also endorsed, particularly for those with **severe feeding difficulties**.
 - Overall consensus is that this must be individualised, depending on the main concern and degree of severity.

Despite the phenotypically heterogeneous nature of ARFID, there is currently no direct evidence that different presentations warrant diverse interventions. Indeed, Dumont et al. (2019) [17], have demonstrated that a flexible CBT approach can be used to treat ARFID with several presentations. Of course, we will only be able to recognise whether different methods are necessary when we know more about the nature of this heterogeneity and begin to test patient responses.

There are several other worthwhile directions for further research including an investigation into ARFID's psychiatric comorbidity, since it has been found to co-occur with various other diagnoses such as generalised anxiety disorder, obsessive compulsive disorder and autism.

3.6 Limitations

- 1) Included studies were of low quality (mainly case studies) with small sample sizes. Further research will need to focus on larger RCT's which use consistent population characteristics and outcome measures.
- 2) There is a wealth of literature relating to sub-clinical restrictive eating behaviours which are symptomatically similar to ARFID, as well as studies pre-dating the introduction of ARFID which would likely provide valuable treatment options for the disorder.

4. Intensive Multidisciplinary Intervention for Paediatric Feeding Disorders

A systematic review conducted by Sharp et al (2017) [28] investigated the medical literature regarding treatment of paediatric feeding disorders at inpatient and day treatment programs. The authors summarise treatment models and outcome measures, and evaluate the evidence with the use of both descriptive and meta-analytic procedures. The sample characteristics (Table 3) and treatment settings and interventions characteristics (Table 4) are summarised below.

4.1 Summary of results

4.1.1 Treatment settings and approach to intervention

- 11 included studies (2 RCT and 9 Non Randomised Studies)
- Collectively the studies include 593 participants (age range 15.7-48 months; 314 boys and 279 girls)
- Treatment for feeding tube dependence (n = 535; 90.2%), liquid formula to meet nutritional needs (n= 22; 3.7%), remaining 36 (6.1%) subjects had various feeding problems but were not tube or formula dependent.
- 8 studies delivered treatment in inpatient facility and 3 in day treatment program and 1 within both settings
- Multiple treatment interventions
 - **Behavioural intervention:** positive reinforcement of appropriate mealtime behaviours, bite persistence (aka, contingency contacting, escape extinction), and/or stimulus fading—represented the most common treatment approach
 - **Oral motor exercises** aimed to decrease tactile hypersensitivity and/or increasing the range, strength and control of the lips, cheeks, jaw and tongue
 - **Tube weaning:** restriction and then reduction
 - **Nutritional intervention:** calculation of energy needs, monitored hydration, adjust tube feeds, tracking of advances
 - All studies involved care givers in treatment

-No study, however, provided specific data on caregivers' acceptance, mastery, and adoption of treatment strategies

4.1.2 Treatment outcomes

- 43% to 100% (Mean 69.8% [SD 21.6%]) of patients were weaned from enteral feeding tubes across the 8 studies that reported this outcome.
- Six studies reported improvement in oral consumption during meals, ranging from 38% to 100% (Mean 74.5 [SD 21.5]) following intervention.
- 36% of studies reported additional gains at follow up, however, 27% reported resumption of tube feeding
- Four studies that included behavioural intervention without tube weaning reported stabilization or improvement in weight.
- The 6 studies that involved tube weaning as a primary treatment component reported weight loss at discharge. Of these, 4 reported on the percentage of weight loss, which ranged from 4% to 9.2%.
- Dependence on enteral feeds was eliminated in 71% of children at discharge. When documented, these benefits appear to persist, with 80% of patients tube-free at follow-up.



Table 3: Summary of sample characteristics

	Study											Total (%)*	
	Brown et al ¹³	Byars et al ¹⁴	Clawson et al ¹⁵	Cornwell et al ¹⁶	Greer et al ¹⁷	Kindermann et al ¹⁸	Hartdorff et al ²⁴	Sharp et al ²⁵	Silverman et al ¹⁹	Trabi et al ²⁰	Williams et al ²¹		
Institution	Children's Hospital of Orange County	Cincinnati Children's Hospital Medical Center	Children's Hospital	Our Children's House at Baylor	Kennedy Krieger Institute	Emma Children's Hospital	Emma Children's Hospital	Marcus Autism Center	Children's Hospital of Wisconsin	Medical Univeristy of Graz	Penn State Hershey Medical Center		
Location	Orange, CA	Cincinnati, OH	Richmond, VA	Dallas, TX	Baltimore, MD	Amsterdam, The Netherlands	Amsterdam, The Netherlands	Atlanta, GA	Milwaukee, WI	Graz, Austria	Hershey, PA		
Design	NRS	NRS	NRS	NRS	NRS	NRS	RCT	RCT	NRS	NRS	NRS		
Sample size	30	9	8	40	121	10	21	10	77	221	46	593	
Sex, n (%)													
Male	18 (60)	5 (55)	4 (50)	20 (50)	71 (58.7)	3 (30)	10 (48)	5 (50)	40 (52)	118 (53)	23 (50)	317 (53)	
Female	12 (40)	4 (45)	4 (50)	20 (50)	50 (41.3)	7 (70)	11 (52)	5 (50)	37 (48)	103 (47)	23 (50)	276 (47)	
Age, mo													
Median	–	–	–	–	–	–	–	–	–	–	37	–	
Mean	48	37.2	32	47.88	45.62	15.7	19.7	44.9	54	26.4	–	–	
SD	16.8	14.4	13.92	16.29	29.70	–	5.4	19.2	26.4	18	–	–	
Range	23-84	21.6-66	18-55	22-84	10-162	9-21	–	–	–	4.5-93	16-133	–	
Primary feeding concern													Studies
Tube dependence (n)	X (30)	X (9)	X (4)	X (40)	X (72)	X (10)	X (21)	X (5)	X (77)	X (221)	X (46)	11 (82%)	
Formula dependence (n)					X (17)			X (5)				2 (18%)	
Other/not specified			X (4)		X (32)							1 (9%)	
Mean age of onset, mo	3	11.6	–	–	–	–	–	–	10.8	–	–	–	
Duration problem, mo	30	26.4	–	–	–	13.5	17.5	–	44.4	21	–	–	
Previous intervention reported	X	X	–	–	–	X	X	–	X	X	X	7 (64%)	
Medical concerns, n (%)													Participants
Cardio/pulmonary	9 (30)	4 (44)	5 (63)	3 (8)	–	2 (20)	8 (38)	7 (70)	39 (51)	41 (19)	10 (22)	128 (27)	
Failure to thrive	–	–	6 (75)	–	–	–	–	4 (40)	–	–	19 (41)	29 (47)	
Food allergies	–	–	–	–	–	3 (30)	1 (5)	1 (10)	–	–	7 (15)	12 (14)	
Gastroesophageal reflux	23 (77)	9 (100)	5 (63)	10 (25)	–	1 (20)	3 (14)	6 (60)	–	–	39 (85)	96 (55)	
General GI problem	9 (30)	6 (66)	1 (13)	–	84 (69)	1 (20)	–	–	71 (92)	46 (21)	11 (24)	229 (44)	
Prematurity	17 (57)	–	7 (88)	24 (55)	24 (20)	3 (30)	7 (33)	–	–	78 (36)	6 (13)	142 (31)	
DD/autism/neurologic	10 (33%)	3 (33)	8 (100)	–	21 (17)	–	4 (19)	3 (30)	52 (77)	18 (8.2)	20 (43)	136 (25)	



Table 4: Treatment setting and intervention characteristics

	Study											Total (%)
	Brown et al ¹³	Byars et al ¹⁴	Clawson et al ¹⁵	Cornwell et al ¹⁶	Greer et al ¹⁷	Kindermann et al ¹⁸	Hartdorff et al ²⁴	Sharp et al ²⁵	Silverman et al ¹⁹	Trabi et al ²⁰	Williams et al ²¹	
Setting												
Inpatient	X	X		X	X	X	X		X	X		8 (73)
Day treatment			X		X			X			X	4 (36)
Treatment duration, d												
Mean (SD)	19	11.4	29	46.43	46.8	17	14.4	5	10.9	21.6	24	22.3 (13.7)
Range		5-16		15-80		9-26				2-52	8-45	
Contributing disciplines												
Gastroenterologist/physician	X	X	X	X	X	X	X	X	X	X	X	11 (100)
Nursing/nurse practitioner	X		X	X		X	X	X				6 (55)
Nutrition/dietician	X	X	X	X	X	X	X	X	X	X	X	11 (100)
Occupational therapist	X			X	X			X		X	X	6 (55)
Psychologist	X	X	X	X	X	X	X	X	X	X	X	11 (100)
Speech-language pathologist	X			X	X	X	X	X	X	X	X	9 (82)
Social worker	X							X				2 (18)
Intervention mechanism(s)												
Behavioral intervention	X	X	X	X	X			X	X		X	8 (73)
Nutrition education	X									X		2 (18)
Oral-motor exercises	X		X	X	X					X		5 (45)
Tube weaning	X	X				X	X		X	X		6 (55)
Behavioral elements												
Contingency contacting/extinction	X	X	X	X				X	X		X	7 (64)
Differential attention	X		X									2 (18)
Negative reinforcement		X		X							X	3 (27)
Positive reinforcement	X	X	X	X		X†	X†	X	X		X	9 (82)
Response cost	X		X								X	3 (27)
Shaping/fading	X	X						X	X		X	5 (45)
Not specified/used					X‡					X§		2 (18)
Caregiver training	X	X	X	X	X	X	X	X	X	X	X	11 (100)

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One further study which investigated intensive multi-disciplinary behavioural treatment for feeding disorders has been published since the systematic review by Sharp et al (2017) [28] (Table 5).

Table 5: Additional published intensive multi-disciplinary treatments for feeding disorders

Author (year) and country	Study aim	Methodology and sample	Symptoms/presentation	Treatment	Outcome
Seiverling et al. [29] 2019 USA	Examine the effects of intensive interdisciplinary behavioural treatment on 11 feeding outcomes	Retrospective chart review 52 children (ASD = 16, other special needs = 19, NAD = 17)	All children exhibited problem behaviours during mealtimes which prevented advancement in diet variety and/or consumption. All cleared of feeding safety concerns	-Attended day treatment facility between 8.15-3.00 Mon-Fri - positive reinforcement for acceptance of target foods -stimulus fading to increase bite sizes -escape extinction (non-removal of spoon) contingent upon inappropriate mealtime behaviour - dietitian, paediatric nurse practitioner, and gastroenterologist provided nutritional and medical monitoring	-Improvements in all outcomes except fruit acceptance -Intervention length 2-8 weeks -Follow up lacked specifics around improvements -small sample restricts generalisability and lack of control group

4.2 Discussion points

There are positive outcomes associated with day treatment and inpatient programs which utilise a multi-disciplinary approach to severe paediatric feeding problems. All studies reported improvements in consumption following interventions.

The below considerations should be taken into account when utilising this systematic review as evidence for the treatment of ARFID.

- 9) 9/11 included studies were published before the introduction of ARFID as a diagnosis in the DSM-5
 - Dependence on enteral feeding or oral nutrition was used as a substitute for an ARFID diagnosis.

-This means results cannot be generalised to the broader ARFID population as we cannot be certain how many included participants will clinically have an ARFID diagnosis.

- 10) Majority of included studies were of low quality (non-randomised)
- 11) 82% tube dependence – more severe form of feeding disorder
- 12) Considerable heterogeneity
 - Outcome measures highly variable. Only tube weaning could be included in meta-analysis
 - Variable primary feeding and medical concerns (25% with ASD/developmental delay/neurologic)
 - Majority of settings were inpatient (8/11)
- 13) Variable treatment duration Mean =22.3 days (SD 13.7)
- 14) Every study included a gastroenterologist/physician, nutritionist/dietician and psychologist
- 15) Behavioural intervention was most commonly used (73%), however, only two studies used the intervention in isolation.
- 16) Of those studies that utilised behavioural intervention, 82% used positive reinforcement, 64% contingency contacting/extinction and 45% used fading/shaping.

The authors note that *“available evidence suggests intensive multidisciplinary treatment likely holds benefits for children with severe feeding difficulties, particularly in cases involving complex medical histories that cannot be effectively managed in an outpatient setting.”*

Current literature involves notable differences in the sequence, timing, and volume of tube feed reduction. Greater specificity regarding the target(s) of intervention and discharge criteria is recommended.

More consistent reporting of follow-up data also is needed to assess the durability of treatment over time. Improved measurement also should entail better characterisation of patients at baseline, including clarity regarding medical and/or behavioural barriers to

achieving oral intake. Given the need for better patient characterization, more uniformity in outcome measurement and unanswered questions on the necessary components of treatment, these 11 studies prohibit definitive conclusions regarding optimal models of care. More systematic evaluation of different treatment approaches and adjuncts to behavioural intervention and/or tube weaning is warranted.

5. Individual Behavioural and Sensory Interventions for Children with Feeding Difficulties

Despite the high prevalence of feeding difficulties in children with ASD, and the implications for short- and long-term health, research regarding intervention for feeding difficulties in this group is scant. It has been shown that clinicians most commonly use therapy approaches based on either operant conditioning (behavioural intervention) or systematic desensitization (sensory intervention) in their treatment for children with ASD and feeding difficulties [30].

Across therapy interventions, those based on operant conditioning currently have the strongest evidence base. However, the majority of existing behavioural research depicting effective specific feeding treatment protocols consist of single case studies or small sample sizes. Operant conditioning interventions use an externally driven 'top-down' approach to prompt the child to perform a desired behaviour, often in conjunction with chaining and/or shaping, and then provide a response contingent on that behaviour. Systematic desensitization is an internally driven 'bottom-up' approach that involves exposure to a feared stimulus (i.e. food) in the presence of relaxation or play activities. Systematic desensitization is also commonly used in the treatment of feeding difficulties but seldom reported in the literature. Table 6 summarises recent systematic reviews that investigate the efficacy of behavioural and sensory interventions for feeding disorders (primarily in those with ASD). Additionally, several recently published RCT's relating to the comparison of operant conditioning and systematic desensitisation are also presented.

Table 6: Summary of literature investigating behavioural and sensory interventions for feeding disorders

Author (year) and country	Study aim	Methodology and sample	Symptoms/ presentation	Treatment	Outcome
Behavioural techniques					
Silbaugh et al. 2017 [31]	Evaluate the certainty of the evidence to guide the evidence-based practice of ABA in the treatment of packing	Systematic review of single-subject designs 7 included studies (6 clinical settings and 1 school) Reflux, failure to thrive, autism, development delay, gastronomy tube	5/7 studies didn't report patient symptoms. One child packed new or non-preferred foods and one held foods until they dissolved	Antecedent manipulations = 5 (71%) Consequence manipulations = 6 (86%)	All studies demonstrated positive outcomes. However, they were rated as 'suggestive' (lowest level) evidence. -All studies were published in only 2 journals - Further treatment replications are required to enable the evaluation of the certainty of the evidence.
Ledford et el. 2018 [32]	(a) What types of interventions have researchers evaluated for individuals with ASD related to mealtime behaviours, and what types of dependent variables have they addressed? Who implemented study procedures, and in what settings were the studies	Systematic review All study type included if there was a comparison condition included All ASD participants Sixty-five articles or manuscripts with 202 designs	-Highly selective eating (i.e., eating fewer than 15 foods; 46%) -Problematic mealtime behaviours such as aggression or disruption (38%) -Unspecified selectivity (29%)	Average of 2.87 components per study Contingent rewards (n = 145) Non-removal of spoon (n =68) Stimulus shaping or fading (n = 63) Re-presentation (n = 62) Response prompting (n = 60) Non-contingent rewards (n = 38) Response shaping (n = 41) Simultaneous presentation (n = 23) Scheduling or restricting food or liquid (n = 17) Behavioural momentum (n = 13) Visual supports (n = 9) Provision of negative consequences (n = 10)	- Clinics (outpatient and inpatient) = 88; Homes = 71; Schools = 24; Residential settings = 9; Unspecified = 9 - Total success rate was 75% for studies addressing acceptance, 45% for problematic mealtime behaviour, and 54% for rumination or vomiting. - Interventions lasted between 2 and 220 sessions (mean = 31) - 50 studies included a maintenance measure, only 4 (8%) reported that outcomes were not maintained.

	conducted? (c) What were the outcomes, and are they different across independent and dependent variable types, settings, or implementers?		-Rumination or vomiting (18%).	Choice (n = 8) Sensory-based Antecedents (n = 5).	- More research is needed to determine for whom and under what conditions feeding interventions are effective, especially for problematic mealtime behaviours. - Little information is available regarding the generalization and maintenance of treatment outcomes.
Silbaugh et al. 2016 [33]	(a) summarise study and participant characteristics of behaviour analytic treatments for food selectivity in children with ASD, (b) evaluate methodological rigor and evidence quality using current standards for evidence based practice in special education	Systematic review Inclusion criteria: at least 1 participants with ASD, Asperger's disorder, pervasive developmental disorder (b) evaluated a behavioural intervention of food sensitivity; and (c) used a single-subject design including graphed data to allow for visual analysis of treatment effects and outcomes.	-Disordered Feeding -Mealtime challenging behaviour	27 studies (96 %) evaluated a treatment consisting of two or more components. One study (4 %) evaluated a treatment component (simultaneous presentation) in isolation. -Differential reinforcements of target feeding behaviour with high preferred food (n = 14, 45 %) -Escape extinction (EE) including non-removal of the spoon (n = 12, 39 %) -Contingent praise (n = 27, 87 %) -Rules (n = 10; 32 %), -Simultaneous presentation (n = 7; 23 %), - Stimulus fading (n = 7; 23 %) -Demand fading (n = 7; 23 %) -Differential reinforcement of feeding responses with non-food reinforcers (n = 9; 29%).	-Current synthesis yielded no information with respect to whether children who have received treatment had nutritional deficiencies or improved their nutrition status following treatment. -Lack of formal outcome measures. Generally use qualitative rather than quantitative approaches -Behaviour analytic treatments for food selectivity appear to produce relatively better improvements in disordered feeding than in mealtime challenging behaviour -Most studies (86 %) combined two or more treatment components, including praise, making it difficult to conclude with certainty in many cases precisely which treatment components were responsible for changes in target behaviours -Standards to determine evidence based practice found that behaviour analytic treatments of food selectivity for children with ASD were classified as having insufficient evidence .

<p>Marshall et al. 2014 [34]</p>	<p>To assist clinicians in decision-making regarding early intervention for children with ASD and feeding difficulties, and to direct further research.</p>	<p>Systematic Review</p> <p>-experimental design was used to investigate treatment outcomes (control group, within group designs, or single-case based)</p> <p>Children with ASD aged 0-6 years</p>	<p>Unclear. Inclusion criteria states 'difficulties relating to eating' 'food selectivity;</p>	<p>Intervention was predominantly provided in an intensive format (multiple times daily) ($n = 10$, 43%), parents were the therapy agents in at least one treatment stage in nearly half of the studies ($n = 11$, 48%), and some component of treatment was completed in the child's home in 61% of the studies ($n = 14$).</p> <p>Intervention feature</p> <ul style="list-style-type: none"> -Antecedent -Response -Consequence -Reinforcement -Punishment -Non-removal of spoon -Thinning reinforcement -Non-contingent reinforcement -Escape as a negative punishment 	<p>-Increasing desirable behaviours: consistent positive effect, mean across all studies being 0.69 (95% CI 0.60 to 0.79)</p> <p>-Undesirable behaviours: mean for these studies being 0.39 (95% CI 0.18 to 0.60).</p> <p>-Trend towards lower effect size in studies where more sessions were provided</p> <p>-Trends towards more successful intervention outcomes where parents undertaking intervention in their home environments</p> <p>-Intensity of intervention provided (e.g. multiple times per day) appeared to have no impact on effect size</p>
<p>Comparison of behavioural and sensory techniques</p>					
<p>Chawner et al. 2019 [35]</p>	<p>Identify interventions used with developmentally disordered populations and to assess their effectiveness in promoting healthy eating behaviours including increasing dietary variety</p>	<p>Systematic review</p> <p>30 case studies, 3 pre-post intervention design, 1 cross-sectional, 1 retrospective chart review</p> <p>Excluded all eating disorders including ARFID</p>	<p>Symptoms/presentations of included participants not reported.</p>	<p>Operant conditioning – escape extinction, non-removal of spoon, physical guidance, differential reinforcement or alternative behaviour, non-contingent reinforcement, lag schedules</p> <p>Based on exposure – systematic desensitisation, stimulus/texture and fading, simultaneous presentation, modelling, high probability sequences, choice of foods, access to preferred food</p> <p>Familial and environmental methods – psychoeducation, parental training,</p>	<p>34/36 reported positive or effective results</p> <p>Techniques from all groups have been reported to be effective (although environmental interventions were only effective when combined with family interventions) for increasing healthy eating of an individual, Case-by-case basis, by increasing the number of new foods eaten, the percentage of bites accepted during a meal and the amount (weight) of new foods that have been consumed.</p> <p>Authors state "Although escape extinction techniques have been consistently reported as most effective, exposure and reinforcement</p>

		<p>19 clinical, 9 home setting, 5 school</p> <p>Majority ASD, ID, pervasive development delay, down syndrome, ADHD</p>		<p>mealtime plans, positive behaviour support, environmental interventions</p>	<p>techniques should be tried before escape extinction and physical guidance strategies due to ethical reasons and to avoid the possibility of adverse side effects</p> <p>-No follow up to determine long term effectiveness</p> <p>- Overall, the evidence was not sufficiently robust to determine the effectiveness of these strategies on a population level.</p>
<p>Reinoso et al. (2018) [36]</p>	<p>What is the evidence of the effectiveness of Sequential Oral Sensory (SOS), Sensory Integration (SI), and (Differential Reinforcement of Alternative Behaviour) DRA interventions for food selectivity and sensitivity in children with ASD?</p>	<p>Systematic review (cohort studies to case series)</p> <p>Unclear – can only assume all included studies only investigated ASD</p>	<p>Symptoms/presentations of included participants not reported</p> <p>Ages ranged from 3 months to 14 years</p>	<p>Outcomes measured</p> <p>SOS: progression in feeding developmental milestones, increased repertoire of foods, mealtime behaviour and positive sensory responses, self-feeding, food rejection</p> <p>SI: mealtime behaviour, increased repertoire of foods</p> <p>DRA: self-feeding, mealtime behaviours, intake of non-preferred foods, food refusal, destructive behaviour</p>	<p>SOS: Several studies have demonstrated promising results. One included study reported no statistically significant improvements, however, it was a crossover design that may have confounded results due to SOS’s impact being exponentially greater with longer duration of treatment.</p> <p>SI: Results were mixed and inconclusive. Possibly best as an adjunct intervention.</p> <p>DRA: far more research published on DRA as compared to SOS and SI. Research confirms the short-term benefits of this approach, with limited long-term validity. DRA is supported for food selectivity.</p> <p>DRA has the most consistent findings in support of its use for food selectivity. SOS is highly recommended because it addresses sensory-based and behaviour-based aversions; whereas SI addresses sensory-based and DRA addresses behaviour-based. Further research is required in the field of SOS to improve its evidence base.</p>

Marshall et al. (2014) [37]	Determine whether intervention across 2 therapy arms (Operant conditioning vs Systematic Desensitisation) had an impact on increasing dietary variety and quality and decreasing the frequency of undesirable mealtime behaviours in children with feeding difficulties	<p>RCT</p> <p>Feeding difficulties in children with an ASD diagnosis and those considered non-medically complex (never received treatment for a medical condition)</p> <p>78 eligible participants</p>	<p>Food selectivity by type (<10 foods across each food group: fruits/vegetables, proteins, carbohydrates)</p> <p>Food selectivity by texture (eg, only consuming purees)</p> <p>Mealtimes averaging >30 minutes, and/or clinically significant difficult mealtime behaviours that were having an impact on parental stress.</p>	<p>-10 sessions consisting of 30-60 minutes (either 10 in one week or weekly over 10 weeks)</p> <p>-Systematic desensitisation (SysD): “Bottom up” modelling and play based therapy</p> <p>-Operant conditioning (OC): Top-down prompt and reward therapy</p> <p>Number of foods offered, short and long term goals, parent involvement and generalisability were the same across both treatment interventions</p>	<p>No different in efficacy of interventions</p> <p>Total number of foods consumed by OC group was clinically greater but not statistically significant</p> <p>No differences observed between etiological groups or intensity (weekly vs intensive intervention)</p> <p>3 month follow up showed continued improvements however treatment groups were not separated.</p>



<p>Marshall et al. (2018) [38]</p>	<p>To examine the outcomes of therapy intervention for medically complex (MC) versus Non MC participants overall, OC versus SysD intervention, and intensive versus weekly therapy intensity dose; and to examine the parent satisfaction following access to a feeding therapy program.</p>	<p>RCT</p> <p>MC: premature, cardiac, respiratory, genetic, neurological, or gastrointestinal conditions; or children with a history of cancer</p> <p>Non MC as above</p> <p>98 eligible participants and 64 completed intervention</p>	<p>As above</p>	<p>As above</p>	<p>Statistically and clinically significant favourable changes to outcome measures for children receiving either intervention were observed.</p> <p>When delivered to a protocol, with consideration of the sensory motor skills of the child, and with the inclusion of parent training, OC or SysD approaches can be successful forms of treatment for feeding difficulties.</p> <p>Parents of children in the MC arm were significantly more likely to elect for intensive intervention than weekly (<i>P</i> 0.02).</p>
<p>Galpin et al. (2018) [39]</p>	<p>To examine the impact of a sensory based intervention to address food selectivity in autistic pupils that could be delivered in a school setting by teaching staff</p>	<p>Repeated-measures within-subject design</p> <p>19 children (3 girls and 16 boys) with ASD who ranged in age from 4 years 10 months to 10 years 7 months (M = 6 years; 5 months; SD = 1;7)</p>	<p>No specific eating or feeding difficulties noted All children had the requisite oral-motor skills to eat table food and had no physical complications, such as dysphagia.</p>	<p>“Sensory Snack Time”: systematic desensitization through the sequential presentation of foods</p> <p>A range of 52 different foods, three liquids and five sauces categorized based upon their texture and food group was made available to pupils during the 12 weeks of Sensory Snack Time sessions, with 4–8 foods available during each session</p>	<p>There were significant improvements in food selectivity score (<i>P</i> <0.001), food refusal (<i>P</i> 0.005) and number of foods tried (<i>P</i> 0.003) post-intervention</p> <p>Results indicated that pupils ate a wider variety of foods and displayed significantly reduced food selectivity, distressed mealtime behaviours, and food refusal following the 12-week intervention</p> <p>Further research is necessary to qualify the precise impact the intervention had and to examine the potential for the intervention to be generalized to main meals and different settings, such as pupils’ homes</p>



The below article was not included in this synthesis as there was significant overlap of included studies with more recent reviews conducted by Silbaugh et al (2016), (2017), Marshall et al (2014), Ledford et al (2018) and Chawner et al (2019)

Sharp WG, Jaquess DL, Morton JF, Herzinger CV. Pediatric feeding disorders: A quantitative synthesis of treatment outcomes. Clinical child and family psychology review. 2010 Dec 1;13(4):348-65.

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5.1 Discussion points

All systematic reviews investigating behavioural interventions concluded that the level of evidence was low or 'suggestive'. This is due to small sample sizes, case study designs and inconsistent outcome measures.

Silbaugh et al. 2016 [33] concluded that *“standards to determine evidence based practice found that behaviour analytic treatments of food selectivity for children with ASD were classified as having **insufficient evidence.**”*

There was little information available regarding the generalisation and maintenance/follow up of treatment outcomes.

The intensity of intervention provided (e.g. multiple times per day) appeared to have no impact. There was a trends towards more successful intervention outcomes where parents undertaking intervention in their home environments

Further research using standardised protocols and randomised study designs are required to enable the evaluation of the certainty of the evidence. This will enable researchers and clinicians to determine for whom and under what conditions feeding interventions are effective, especially for problematic mealtime behaviours.

Systematic reviews comparing sensory and behaviour interventions found that techniques from all groups have been reported to be effective (although environmental interventions were only effective when combined with family interventions) for increasing healthy eating of an individual (increasing the number of new foods eaten, the percentage of bites accepted during a meal and the amount (weight) of new foods).

Chawner et al. (2019) [35] concluded that *“Although escape extinction techniques have been consistently reported as most effective, **exposure and reinforcement techniques should be tried before escape extinction and physical guidance strategies due to ethical reasons** and to avoid the possibility of adverse side effects*

This was reiterated by Reinoso et al. (2018) [36] who stated that Sensory interventions are highly recommended because they address sensory-based and behaviour-based aversions