

# Efficacy of interventions to improve feeding difficulties in children with autism spectrum disorders: a systematic review and meta-analysis

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## Abstract

**Background** Feeding difficulties are relatively common in children with autism spectrum disorders (ASD), but current evidence for their treatment is limited. This review systematically identifies, reviews and analyses the evidence for intervention in young children with ASD and feeding difficulties.

**Methods** A comprehensive search strategy was used to identify studies from January 2000 to October 2013. Studies were included if they described interventions where the goal was to increase desirable eating behaviours or decrease undesirable eating behaviours using an experimental design, including single-subject research methodology. Studies were reviewed for descriptive information, and research quality was appraised using a formal checklist. Individual study findings were compared using Improvement Rate Difference (IRD), a method for calculating effect size in single-subject research.

**Results** Overall, 23 papers were included. All studies reviewed had five or fewer participants, and reported on operant conditioning style intervention approaches, where the child is prompted to perform an action, and receives a contingent response. Where quality measures were not met, it was primarily due to lack of detail provided for the purposes of replication, or failure to meet social validity criteria. Meta-analysis indicated a medium-large effect size [mean = 0.69, 95% confidence interval (CI) 0.60 to 0.79] when the outcome measured was an increase in desirable behaviours (e.g. consuming food), but a small-negligible effect size (mean = 0.39, 95% CI 0.18 to 0.60) when the outcome measured was a decrease in undesirable mealtime behaviours (e.g. tantrums). Only a small proportion of studies reported outcomes in terms of increased dietary variety rather than volume of food consumed.

**Conclusions** The reviewed literature consisted primarily of low-level evidence. Favourable intervention outcomes were observed in terms of increasing volume, but not necessarily variety of foods consumed in young children with ASD and feeding difficulties. Further research in the form of prospective randomized trials to further demonstrate experimental effect in this area is required.

## Keywords

autism spectrum disorder, eating, feeding difficulties, feeding disorders, interventions, treatment

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## Introduction

Feeding difficulties have been identified as occurring in up to 25% of typically developing children (Lindberg *et al.* 1991), with behaviours such as picky eating, fussy eating or food neophobia (fear of new foods) often described (Cashdan 1998). For children with autism spectrum disorder (ASD), however, this incidence has been observed to be as high as 89% (Ledford & Gast 2006). The nature of feeding difficulties in children with ASD has been described as including extreme food neophobia, restricted dietary variety, food selectivity by texture and a propensity towards being overweight (Marshall *et al.* 2014).

Both short- and long-term health consequences have been documented for children experiencing feeding difficulties and restricted dietary variety. In the short term, where limited dietary variety leads to reliance on energy-sparse foods, inadequate energy consumption, weight loss or failure to thrive may occur (Bolte *et al.* 2002; Keen 2008). Conversely, if there is an over-reliance on energy-rich, but nutrient-poor foods, this may result in weight gain, such that a child is overweight, but malnourished (Ho *et al.* 1997; Matson *et al.* 2009; Xiong *et al.* 2009). Medical complications, such as gastrointestinal discomfort (Bosaeus 2004) and iron deficiency anaemia (Latif *et al.* 2002), may also arise from consuming a restricted diet. Difficult behaviours at mealtimes and concern regarding poor intake may also contribute to increased parental stress (Greer *et al.* 2008). Long-term consequences of poor dietary variety habits in childhood include increased risk of overweight as an adult (Kelder *et al.* 1994), which has resultant implications for the development of diseases of later life (Lucas 2005; Rimmer *et al.* 2010).

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. A recent survey of practice indicated that clinicians most commonly use therapy approaches based on either operant conditioning or systematic desensitization in their treatment for children with ASD and feeding difficulties (Marshall *et al.* 2013). Across therapy interventions, those based on operant conditioning currently have the strongest evidence base (Kodak & Piazza 2008; Sharp *et al.* 2010). Interventions using this externally driven 'top-down' approach 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 (Marshall *et al.* 2013), but seldom reported in the literature. Of concern, clinicians working with children with ASD and feeding difficulties have indicated low levels of confidence in their knowledge of the area and perceived therapy success (Marshall *et al.* 2013), which suggests a need for research to support the development of practice guidelines.

An examination of the literature on interventions for children with ASD and feeding difficulties reveals that few systematic reviews have been undertaken. One review identified nine intervention studies over a 10-year period, and concluded that therapy was effective overall in the treatment of feeding problems in children with ASD, despite there being a wide variety of therapy approaches used (Ledford & Gast 2006). In their systematic review of 25 studies, Mari-Bauset and colleagues (2013) reported improvements in energy intake per meal and weight gain in response to behavioural interventions, but also concluded that the quality of research reviewed was weak. Other reviews to date have not been systematic in nature, instead superficially describing a few selected studies or common interventions used (Kodak & Piazza 2008; Matson & Fodstad 2008; Williams & Seiverling 2010).

The current paper systematically identifies, reviews and analyses the evidence for early interventions for children less than 6 years of age with ASD and feeding difficulties. This review was undertaken to answer the following clinical question: In young children with ASD and feeding difficulties, does early therapy intervention result in improvement of mealtime intake and mealtime behaviours? Our first aim was to review the quality of identified studies. Where possible, we extracted data based on the primary outcomes of increased volume and variety of intake (increasing desirable mealtime behaviours). We also collected information on the secondary outcome of reduction of inappropriate mealtime behaviours. Our second aim was to collect and report on information regarding dose of intervention, implementation of parent training for generalization, and length of time between intervention and post-treatment measures. The overall goals of this review were to assist clinicians in decision-making regarding early intervention for children with ASD and feeding difficulties, and to direct further research.

## Methods

### Selection criteria

To be included in this systematic review, studies had to meet the following criteria: (1) they included children aged 0–6 years with a diagnosis of ASD; (2) interventions delivered were

intended to improve intake (volume of food and/or variety of foods consumed) and/or eating behaviours; (3) an experimental design was used to investigate treatment outcomes, including the use of a control group within group designs, or single-case-based experimental research methodology; and (4) studies were published in English in peer-reviewed journals between January 2000 and October 2013.

Studies were excluded from review if they: (1) reported interventions that did not primarily focus on or address difficulties related to eating; (2) reported pharmaceutical interventions; (3) reported interventions where the focus was on the manipulation of diet to improve behaviour; (4) provided intervention for feeding behaviour that was not disruptive to mealtimes or intake (e.g. pica); and (5) did not include a control condition (e.g. case studies where experimental control was not demonstrated).

### Search strategy

A comprehensive search was conducted on 10 October, 2013 using the following databases: PubMed (2000–October 2013), CINAHL (2000–October 2013), PsycINFO (2000–October 2013), the Cochrane Database of Systematic Reviews, ERIC (2000–October 2013), speechBITE and OTseeker. The year 2000 was selected as the initial year of review, as criteria for diagnosis of ASD were revised according to the Diagnostic and Statistical Manual for Mental Disorders (DSM-IV-TR) during this year (American Psychiatric Association 2000).

The search strategy included the following Medical Subject Headings (MeSH) headings or keywords: (1) autism spectrum disorder *or* autism *or* autistic *or* Asperger\* *or* pervasive developmental disorder; *and* (2) feeding and eating disorders of childhood *or* feeding behaviour *or* feeding difficulties *or* feeding disorder *or* mealtime *or* food selectivity *or* picky eat\* *or* eating habits; *and* (3) behavior modification *or* operant conditioning *or* systematic desensitization *or* parent education *or* parent training *or* intervention *or* nonremoval *or* reinforcement *or* punishment. Reference lists of identified papers were also searched for additional references.

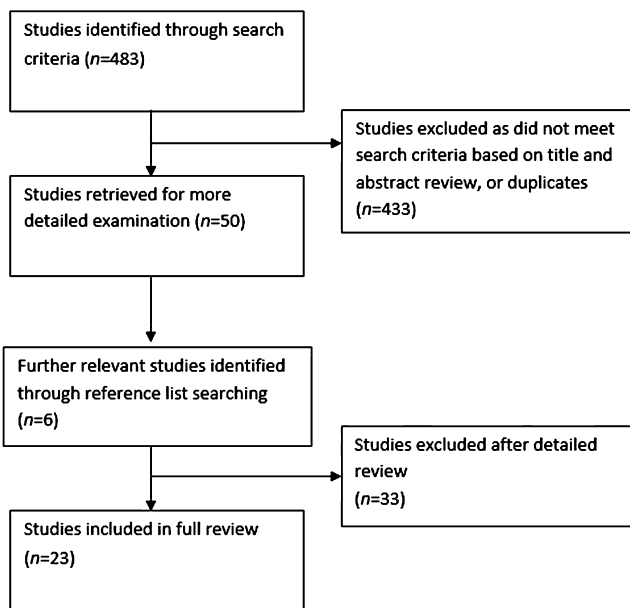
Two authors (JM, PD) reviewed all abstracts for suitability. Abstracts of final studies for inclusion were reviewed by four authors (JM, PD, RH, JZ). Two authors (JM, PD) reviewed the full text of these studies. Quality data were independently appraised and rated by two authors (JM, JZ). Study effectiveness data were extracted independently by two authors (JM, RW). Where there were differences, a third author was consulted (PD), and consensus reached.

### Analysis

Descriptive data regarding level of evidence, goals of study, intervention type provided, duration and outcomes were collated. Quality appraisal of the included studies was completed using a tool for assessing quality indicators within single-subject research (Horner *et al.* 2005). Each appraisal criterion was comprised of multiple components. A paper met each criterion if it addressed all components. The authors made the following assumptions across some of the components where there was opportunity for subjective interpretation. Under *Description of participants and settings*: for (1) *Participants are described with sufficient detail to allow others to select individuals with similar characteristics*, the criterion was met if the paper adequately described age, diagnosis, developmental level, medical history and cognitive history; for (2) *The process for selecting participants is described with replicable precision*, the criterion was met if the criteria for including the participant in the study were specified; and for (3) *Critical features of the physical setting are described with sufficient precision to allow replication*, the criterion was met if room set-up, utensils and positioning for the participant were detailed. In the *Social Validity* section, (1) *The magnitude of change in the dependent variable resulting from the intervention is socially important* was met if the family completed a favourable social validity questionnaire; and for (2) *Implementation of the independent variable is practical and cost-effective*, the criterion was met if the authors described a means of cost-analysis.

For all included studies where a graphic representation of response to therapy was provided as part of the study's results (typically provided for the purposes of visual analysis), data extraction was completed ( $n = 22$ ). Data analysis was undertaken using Improvement Rate Difference (IRD), a method for calculating effect size in single-subject research (Parker *et al.* 2009). IRD is defined as the difference in improvement between the treatment and baseline phases, and is mathematically equivalent to the widely used *risk difference* (Parker *et al.* 2011). Details for calculating IRD are described in Parker *et al.* (2009). The method of two proportions was used to calculate a 95% confidence interval (CI). Where multiple phases were analysed, results were combined and new IRD and CIs were calculated using the inverse variance weighting method. As a result of difficulties with computing standard error from cells where there were zero values, 0.5 was added to each cell in these cases (Higgins & Green 2011).

There was concern in analysis of these IRD data that some studies did not allow for a suitable 'washout period', where there were multiple shifts between intervention and control



**Figure 1.** Included and excluded studies.

phases. It was felt that this may have impacted the IRD and, therefore, not been representative of the true effects of the intervention provided. While the primary analyses were conducted across all phases to maintain consistency with other reviews in the area (e.g. Ganz *et al.* 2012), as a result of the above concerns, an additional sensitivity analysis was conducted that considered the initial baseline and intervention phases only. Results from these sensitivity analyses may be interpreted as the potential effect to be gained from a single baseline-intervention condition.

## Results

The search strategy identified 483 potential abstracts (Fig. 1). Fifty studies were retrieved for full analysis, and six further studies were identified through reference list searching. Review of the full text of these studies identified 23 studies, which were included for full analysis according to the selection criteria.

### Descriptive information

Descriptive information about the studies included for full review is displayed in Table 1. In most studies, the stated aim was to treat 'food selectivity', suggesting the primary goal of treatment was to increase dietary variety. Despite this, the dependent variable most often described focused on volume

intake (number or percentage of bites consumed of a limited number of foods). Only a small proportion of studies anecdotally reported an increase in the number of foods eaten ( $n = 5$ , 22%), and only two studies used a formal outcome measure to capture this information (Paul *et al.* 2007; Pizzo *et al.* 2009).

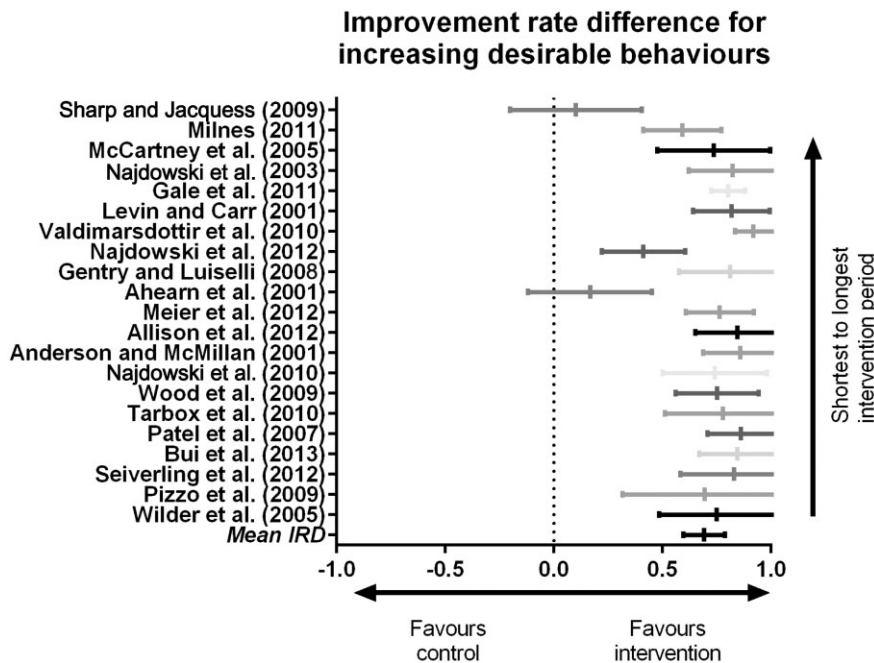
A detailed description of each dependent and independent variable is provided. All studies retrieved for full analysis demonstrated experimental control via baseline and intervention conditions, but were presented as either single cases (12 studies) or small group interventions only (11 studies). Of the studies described, the 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 ( $n = 14$ ). Further details regarding therapy provided, in terms of antecedents and consequences, are presented in Appendix I.

### Quality review

Quality rating scores ranged from 7 to 18 (out of a possible 21) (Table 2). The total agreement score between authors on the quality review tool was 89%. An extended description of scoring decisions is provided in Appendix II. The majority of studies were rated highly on *Dependent Variable*, *Baseline* and *External Validity* criteria. *Description of Participants/Settings* and *Social Validity* were the two criteria that scored poorly. Participants and settings were often partially described, but key details that would allow comparison with other similar participants were omitted (most commonly cognitive ability, and information regarding current diet). Criteria for social validity were not met because no studies reported directly on cost-effectiveness measures, and it was difficult to objectively gauge the full impact of 'social importance'. *Internal validity* was also poorly rated, with demonstrating experimental effect over three different points in time the most common component not addressed. Finally, in the *Independent variable* criteria, only a few studies reported on treatment fidelity or employed a formal system for its measurement.

### Effectiveness data

Improvement Rate Differences representing increase in desirable behaviours and decrease in undesirable behaviours are presented as forest plots in Figs 2 and 3, respectively. With regard to increasing desirable behaviours (typically accepting



**Figure 2.** Improvement Rate Difference (IRD) for increasing desirable behaviours.

bites of food), studies reported a consistent positive effect, with the mean across all studies being 0.69 (95% CI 0.60 to 0.79), which is considered to be a medium-large effect size (Parker *et al.* 2009). This suggests that the intervention provided generally had positive effects in increasing desirable behaviours. Results were less consistent for effectiveness of intervention on undesirable behaviours, with the mean for these studies being 0.39 (95% CI 0.18 to 0.60). This indicates a small or negligible effect size (Parker *et al.* 2009), and suggests that this intervention had minimal impact on improvement of undesirable behaviours according to these measures.

In Figs 2 and 3, studies are organized from shortest intervention time to longest intervention time to allow for comparison. There was a trend towards lower effect size in studies where more sessions were provided, both in increasing desirable and in decreasing undesirable behaviours. Other analyses revealed trends towards more successful intervention outcomes where the therapy providers were the parents undertaking intervention in their home environments. Intensity of intervention provided (e.g. multiple times per day) appeared to have no impact on effect size in these studies (see Appendix III for raw IRD data, and Appendix IV for forest plots comparing different trends).

Sensitivity analysis revealed no significant difference in IRD across single phase data. The mean for increasing desirable behaviours was 0.71 (95% CI 0.61 to 0.82), and the mean for

decreasing undesirable behaviours was 0.44 (95% CI 0.26 to 0.62).

## Discussion

This review presents a quality appraisal of the current literature in the area of intervention efficacy for children with ASD under the age of 6 years with feeding difficulties. It was completed in order to address limitations in the current state of knowledge for this emerging area. All studies reviewed presented small group or single case data only. The quality of the papers reviewed was variable, with many failing to meet internal or social validity criteria, or providing inadequate information for replication. Meta-analysis through use of the IRD method suggested a favourable response to treatment in terms of increasing desirable eating behaviours (increase in bite acceptance), but an inconsistent response with regard to reduction of undesirable behaviours.

All studies reviewed reported results of operant conditioning interventions. There were no studies which reported on interventions using systematic desensitization, although several used the concepts of chaining (moving from preferred to non-preferred foods), and shaping (performing components of a task to contribute to the overall task e.g. kissing a food). Current lack of focus on the area of systematic desensitization in the literature may be because this is a relatively new area of practice for children with feeding difficulties.



Table 1. Descriptive information

Study	n (ASD)	Age (years)	Programme (independent variable)	Dependent variable (Increase desirable)	Dependent variable (Decrease undesirable)	Agent	Freq.	Total sessions	Location	Follow-up
Ahearn <i>et al.</i> 2001	2 (1)	4	1. NRS + CR (object) 2. PG + CR (object)	% bites accepted	% expulsions % negative voc. % disruptions	Therapist	Multiple/day	50	Inpatient unit	Weekly for 1 month, then once/month Not reported
Allison <i>et al.</i> 2012	2 (1)	3	1. NRS + CR (object and verbal) 2. NRS + NCR (object and verbal)	% bites accepted	# disruptive behaviours/min % duration negative voc.	Therapist	Multiple/day	38	Autism centre	Not reported
Anderson and McMillan 2001	1 (1)	5	NRS + CR (verbal and food)	% bites accepted	% bites interrupted % bites expelled % bites with SIB	Parent	Daily	38	Home	Not reported
Bui <i>et al.</i> 2013	1 (1)	2	NRS + CR (verbal)	% bites accepted	% bites with SIB	Parent	Multiple/day	14	Home	1 × follow-up 1 week post 1 × follow-up 4–5 months post (n = 2)
Gale <i>et al.</i> 2011	3 (3)	3–4	NRS + CR (verbal and food) (+ non-contingent escape)	# bites accepted/20	# trials with disruptive behaviour/20	Parent ABA tutor	Multiple/day	68–73	Home	Not reported
Gentry and Luiselli 2008	1 (1)	4	Random chance game to determine volume + CR (verbal and object) + thinning CR	# bites consumed		Parent	Daily	52	Home	Not reported
Levin and Carr 2001	4 (4)	1 < 6 years	Hunger manipulation +/- CR (food)	# grams consumed	# disruptive behaviours # bites expelled Bite latency	Therapist	5 ×/week	65	School	Not reported
McCartney <i>et al.</i> 2005	4 (2)	5	NRS + CR (verbal and food)/escape + thinning CR	# bites accepted	% 5 s intervals with disruptive behaviours	Therapist Parent		98	Clinic and home	1 × follow-up 1 month (n = 1) and 1 year post (n = 1)
Meier <i>et al.</i> 2012	1 (1)	3	Shaping from preferred to non-preferred foods + CR (verbal)	% bites accepted		Therapist	3–5 ×/week	44	Home	1 × follow-up per food 12 and 15 days post
Milnes 2011	5 (5)	4 < 6 years	Random chance game to determine volume + CR (verbal and object) + thinning CR	# bites accepted	% 10 s intervals with targeted disruptive behaviours	Parent	5 ×/week	100	Home	2 completed full programme Questionnaires 1–3 months post 2, 4, 6 and 12 weeks post
Najdowski <i>et al.</i> 2003	1 (1)	5	NRS + CR (verbal + food)/escape + thinning	# bites accepted		Parent	Daily	~79	Home Restaurant	
Najdowski <i>et al.</i> 2010	3 (2)	2–4	NRS + CR (food) + thinning	% bites accepted	% trials with inappropriate behaviours (collected but not reported)	Parent	2–7 ×/week	36–38	Home	(1) 2 weeks post (2) 2, 4, 6, and 12 weeks post

Table 1. Continued

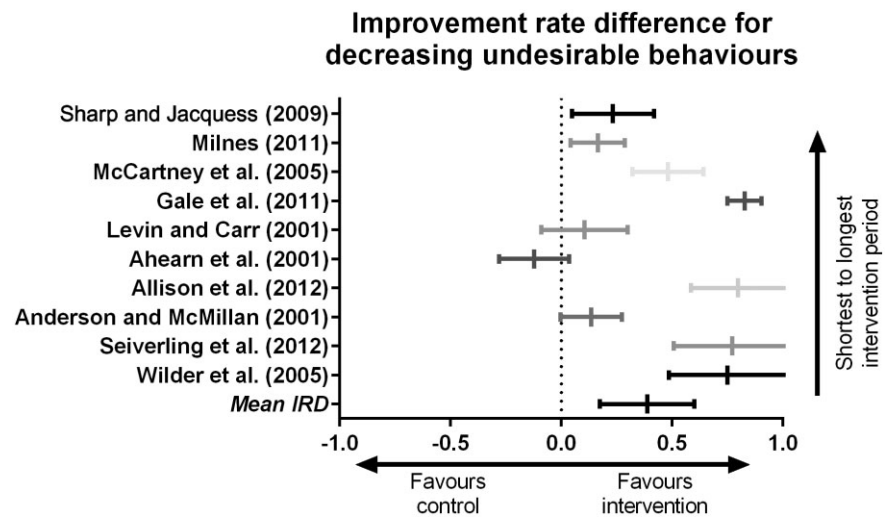
Study	<i>n</i> (ASD)	Age (years)	Programme (independent variable)	Dependent variable		Agent	Freq.	Total sessions	Location	Follow-up
				(Increase desirable)	(Decrease undesirable)					
Najdowski et al. 2012	1 (1)	3	Prompt + CR (verbal + object) + grading (texture) ± simultaneous presentation	% bites accepted (collected but not reported) % mouth clean	% bites accompanied by inappropriate mealtime behaviours (collected but not reported)	Therapist	2–3×/week	~52	Clinic (home)	Not reported
Patel et al. 2007	1 (1)	4	Shaping preferred to non-preferred task + CR (verbal)	% bites (of low-probability foods) accepted		Therapist	Multiple/day	~26	Clinic	1× follow-up 3 months post
Paul et al. 2007	2 (2)	3–5	Taste exposure sessions: NRS + negative reinforcement + grading (volume) Generalization sessions: CR (verbal)	Bite latency # foods where 3 full spoons consumed	% inappropriate mealtime behaviours/total trials	Therapist	Multiple/day	13–15 days	Clinic	1× follow-up 3 months post
Pizzo et al. 2009	3 (1)	4	Replicate procedure by Paul and colleagues (2007) in a shorter time	Bite latency # foods where 3 full spoons consumed	# inappropriate behaviours/meal	Therapist	Multiple/day	4 days	Clinic	1× follow-up 4 weeks post
Seiverling et al. 2012	3 (3) 2 < 6 years	4–5	Taste exposure sessions: NRS + negative reinforcement + fading Probe sessions: CR (verbal)	% bites accepted in <30 s	% bites with inappropriate behaviours	Parent	Multiple/day	10–11 days	Home	1× follow-up/week for 3 weeks
Sharp and Jacques 2009	1 (1)	3	NRS + NCR + grading (volume and texture)	% mouth cleans	% inappropriate mealtime behaviours	Therapist	Multiple/day	~224	Day programme	Not reported
Tarbox et al. 2010	1 (1)	3	Non-removal of the meal + escape	% meal consumed	# expulsions # gags Meal duration	Parent	Multiple/daily?	31	Home	Follow-up at 1, 2, 4 and 9 weeks post Follow-up 25 days after school and 19 days after home setting
Valdimarsdottir et al. 2010	1 (1)	5	Prompt + CR (verbal and object) + thinning	# bites consumed		Therapist Parents Teachers	Daily?	~55	School Home	Not reported
Volkert et al. 2011	2 (1)	5	NRS + CR (verbal) + flipped spoon for redistribution of food	% trials with acceptance % bites accepted	% bites packed	Therapist	Weekly	~26	Clinic	Not reported
Wilder et al. 2005	1 (1)	3	NRS (30 s) + NCR	% trials with acceptance % bites accepted	% 10 s intervals with SIB # escapes	Therapist	2×/week	~12	Clinic	Not reported
Wood et al. 2009	1 (1)	5	Prompting + shaping + CR	% bites accepted		Therapist	4×/week	~32	Home	Not reported

PG, physical guidance; NRS, non-removal of the spoon; CR, contingent reinforcement; NCR, non-contingent reinforcement; EE, escape extinction; voc., vocalizations; SIB, self-injurious behaviours.

**Table 2.** Quality review

Study	Description of participants/ settings	Dependent variable	Independent variable	Baseline	Internal validity	External validity	Social validity	Total (/21)
Ahearn <i>et al.</i> (2001)		✓		✓		✓		14
Allison <i>et al.</i> (2012)		✓	✓	✓				15
Anderson and McMillan (2001)				✓				12
Bui <i>et al.</i> (2013)		✓				✓		14
Gale <i>et al.</i> (2011)		✓	✓	✓		✓		18
Gentry and Luiselli (2008)		✓		✓				14
Levin and Carr (2001)		✓	✓	✓		✓		16
McCartney <i>et al.</i> (2005)	✓		✓	✓	✓	✓		18
Meier <i>et al.</i> (2012)		✓		✓	✓			15
Milnes (2011)		✓	✓	✓		✓		15
Najdowski <i>et al.</i> (2003)		✓		✓		✓		14
Najdowski <i>et al.</i> (2010)		✓		✓		✓		15
Najdowski <i>et al.</i> (2012)		✓		✓		✓		15
Patel <i>et al.</i> (2007)		✓	✓	✓				14
Paul <i>et al.</i> (2007)						✓		7
Pizzo <i>et al.</i> (2009)				✓		✓		13
Seiverling <i>et al.</i> (2012)						✓	✓	13
Sharp and Jacquess (2009)		✓	✓	✓	✓	✓		18
Tarbox <i>et al.</i> (2010)		✓		✓	✓			14
Valdimarsdottir <i>et al.</i> (2010)		✓	✓	✓	✓	✓		16
Volkert <i>et al.</i> (2011)		✓	✓	✓		✓		16
Wood <i>et al.</i> (2009)		✓		✓		✓		14
Wilder <i>et al.</i> (2005)		✓	✓	✓				13

✓ = 100% of criteria met.

**Figure 3.** Improvement Rate Difference (IRD) for decreasing undesirable behaviours.

Given the IRD method used in this meta-analysis is a relatively novel means of comparing outcomes across different studies, results should be interpreted with some care. As a strength, the method does allow for a more objective interpretation of intervention outcomes through calculation of a meaningful effect size and CIs, as opposed to use of visual analysis alone. Potential limitations are present in

measuring change across multiple intervention phases, and where there is a trend towards improvement during both baseline and intervention phases (but no overlap). The method also appears to have some limitations in capturing improvement where performance for the child during baseline is highly variable, or the baseline presented is very short.



The current review has identified a lack of studies with larger sample sizes (>5 participants) where prospective data were gathered, and no studies where intervention was compared in a randomized design. Given the selective reporting of patient outcomes, it is possible that there is publication bias towards papers that report favourable outcomes, and non-reporting of cases where therapy results were not favourable.

Within the studies themselves, there was a great variability in the duration and frequency of interventions, with studies reporting that between 12 and 224 sessions were provided to achieve goals. As can be seen from analysis of the IRD data, longer treatment time and increased intensity did not necessarily equate to better outcomes with regard to increasing desirable or decreasing undesirable behaviours. With respect to the data presented, many studies used number of bites accepted as their primary outcome measure (increasing a desirable eating behaviour). Some studies, however, reported only on proportion of bites accepted, but did not report on how many bites were offered to make up this proportion (e.g. Anderson & McMillan 2001; Allison *et al.* 2012). This has the potential for misrepresenting the true outcomes.

While the IRD method used suggests that results of intervention are generally favourable in terms of increasing desirable eating behaviours, it also highlights inconsistency in reduction of undesirable behaviours. It may be that reduction in undesirable behaviour is a more unstable and difficult phenomenon to measure. Use of IRD may, therefore, be limited in measuring reduction of difficult behaviours, which was generally considered to be a secondary outcome.

The primary aim described across the majority of studies was to treat 'food selectivity', which inherently suggests that the over-arching goal of treatment was to increase dietary variety. Despite this, the number or percentage of bites accepted (i.e. volume intake) was often the only dependent variable measured, and the number of foods consumed (i.e. variety) was only reported anecdotally in a select few studies, and only formally measured in two of these. Analysis of macro- and micro-nutrient intake from a prospective food diary or food variety score information (Cox *et al.* 1997) would be a more meaningful measurement of long-term variety outcomes for these children. On examination, there was a marked difference between studies in the number of foods targeted for therapy, with one being three foods over 44 sessions (Meier *et al.* 2012) and another being 'multiple foods' over 12–15 days (Paul *et al.* 2007) as examples. Effort expenditure in terms of increasing dietary variety by only a small number of foods would be useful to review in gaining a full appreciation of therapy value.

A number of studies ( $n = 9$ , 38%) reported no follow-up for participants. In cases where there was follow-up reported, this was completed a mean of 7.6 weeks after treatment, with the exception of one study that reported follow-up 1 year after treatment (McCartney *et al.* 2005). This was disappointing, given the reported frequent occurrence of relapse of behaviours over time or in different contexts for patients who have been treated with conditioning (Bouton *et al.* 2012). It would be an extension for future research to review long-term outcomes for patients receiving treatment for feeding difficulties.

Of note were the number of studies where the parent was engaged as the therapy facilitator ( $n = 11$ , 48%). Greater improvements in generalization and maintenance have previously been observed where parents are facilitators of therapy (Koegel *et al.* 1982), and many parent-based behavioural studies for children with ASD have identified successful outcomes for participants (Kashinath *et al.* 2006; Jones & Feeley 2010). Analysis of the IRD data identified a trend towards slightly improved feeding therapy outcomes in children where the parent was trained as the therapist. However, this should be interpreted with caution, given the lack of long-term follow-up to allow for consideration of generalization and maintenance.

Most studies did not consider or report on the impact of hunger manipulation as a part of their intervention plan, with only a few exceptions (Ahearn *et al.* 2001; Levin & Carr 2001; Najdowski *et al.* 2010; Gale *et al.* 2011; Seiverling *et al.* 2012). Variable hunger state, either due to lack of hunger (as a result of access to preferred foods before sessions) or due to too much hunger (as a result of rapid weaning from tube-feeding), could present a threat to internal validity. In addition, a number of studies reported limited information regarding the participants involved, particularly with respect to medical history and cognitive level, which made comparison between cases difficult, and would make it difficult to replicate these studies completely.

None of the studies reviewed reported an analysis of cost-effectiveness, which impacted on their quality score for social validity. Analysis of cost-effectiveness (i.e. cost vs. benefits) is an important consideration in managing demands for health care in a competitive market. It could be hypothesized that therapy implemented at home with the parent as the facilitator would be cheaper but perhaps equally effective (thus, more cost-effective) but, without analysis, this assumption is difficult to sustain.

Finally, although quite a few studies involved a secondary rater for a proportion of treatment sessions, only a few employed a formal measure of fidelity to treatment. Consistent

use of fidelity measures has been highlighted as lacking, but essential, in demonstrating intervention effectiveness (Parham *et al.* 2007). Additionally, given the nature of the outcomes collected, it would have been difficult for secondary raters to be blinded, which has implications for bias in data collection and analysis.

## Conclusion

This review presents a novel approach to quality review of a small body of literature regarding early therapy intervention for feeding difficulties in children with ASD. While there were some limitations in the literature reviewed, particularly with regard to the number of single case and small group studies included, the evidence of a positive effect of intervention on increasing dietary intake in terms of volume, not necessarily variety, in young children with ASD was observed. Further research in the form of prospective randomized controlled trials is recommended to fully evaluate the impact of intervention in this group. Use of a well-considered range of outcome measures to capture long-term and wider-ranging impacts, as well as the involvement of a multidisciplinary team, are also advised given the complex nature of feeding difficulties.

### Key messages

- Current literature regarding early intervention for children with ASD and feeding difficulties is limited, and of varying quality.
- Analysis of case study and small-group investigations suggested feeding therapy had a medium-large effect on increasing dietary intake, but a small-negligible effect on decreasing undesirable mealtime behaviours.
- Descriptive information collected from the literature suggested a mismatch between stated intentions (decreasing food selectivity) and measured outcomes (increased intake volume), and a lack of long-term follow-up in many cases.

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**Appendix I**

Features of intervention as described by authors

Study	Goal (therapy agent)	Treatment design	Antecedent	Definition	Consequence	Author definition	Positive	Negative	Reinforcement	Punishment
Ahearn et al. (2001)	Compare physical guidance + CR vs. NRS + CR (therapist)	NRS + CR vs. Physical guidance + CR Session concluded after 20 spoon presentations	Verbal prompt NRS  Physical guidance	'Child's name, open' Spoon remained positioned at lower lip until child opened mouth and allowed spoon to be placed inside If child did not accept bite within 5 s, therapist opened child's mouth by applying constant pressure in front of the mandibular junction of the jaw, and deposited the bite on opening No access to food for at least 1 h before session	CR (object)  Blocking Ignoring Re-presentation  Removal of contingent on stimuli expulsion	Social interaction and access to preferred stimuli presented for >15 s on a fixed 1:1 schedule Therapist prevented child's arms from crossing midline of body Undesirable behaviour ignored Therapist attempted to catch food and re-present, or a new spoon was presented If expulsion occurred during reinforcement period, access to preferred stimuli/attention removed	✓	✓	✓	✓
Allison et al. (2012)	Compare NRS + CR, vs. NRS + NCR (therapist)	NRS + CR vs. NRS + NCR Session concluded after 20 spoon presentations or after 20 min	Hunger manipulation NRS	Bites presented once every 30 s for 5 min. Bite remained at child's mouth until therapist could deposit it	CR (object and verbal)  Blocking	Therapist presented 30 s access to preferred toy + praise and interaction on a fixed 1:1 schedule Problem behaviour blocked if necessary Bites represented until accepted or 20 min passed Preferred item available throughout session Preferred food was offered within 5 s of bite acceptance on a fixed 1:1 schedule Number of bites of NPF required for meal to end increased as success achieved Parents advised to ignore interruptions Meal ended after particular number of NPF bites Verbal praise provided in response to accepted bites on a fixed 1:1 schedule Mother instructed to ignore undesirable behaviours and reintroduce the same spoon if refusal behaviours were engaged in	✓	✓	✓	✓
Anderson and McMillan (2001)	Evaluate NRS + CR (parent)	NRS + CR Session concluded after a pre-determined volume of non-preferred food was consumed (at least one bite)	NRS	Spoon held at child's lips until child opened mouth (i.e. to accept, or yawn/cry)	Representation NCR (object and verbal) CR (verbal and food)  Thinning reinforcement Ignoring Escape	Problem behaviour blocked if necessary Bites represented until accepted or 20 min passed Preferred item available throughout session Preferred food was offered within 5 s of bite acceptance on a fixed 1:1 schedule Number of bites of NPF required for meal to end increased as success achieved Parents advised to ignore interruptions Meal ended after particular number of NPF bites Verbal praise provided in response to accepted bites on a fixed 1:1 schedule Mother instructed to ignore undesirable behaviours and reintroduce the same spoon if refusal behaviours were engaged in	✓	✓	✓	✓
Bui et al. (2013)	Evaluate NRS + CR (parent)	NRS + CR Session concluded after 30 min	NRS	Spoon held at child's mouth until bite accepted	CR (verbal)  Ignoring	Verbal praise provided in response to accepted bites on a fixed 1:1 schedule Mother instructed to ignore undesirable behaviours and reintroduce the same spoon if refusal behaviours were engaged in	✓	✓	✓	✓
Gale et al. (2011)	Compare NRS + escape vs. NRS + CR (parent/ABA tutor)	NRS + escape (BL) vs. NRS + CR (+ non-contingent escape) Session concluded after 20 spoon presentations	NRS Hunger manipulation Escape extinction	Bite remained at child's mouth for 30 s each trial No access to preferred food provided in 30 min prior to intervention Child in restrained seating that did not allow escape from the feeding situation	Non-contingent escape CR (object and food) Blocking	Spoon briefly removed after 30 s trial; meal ended after 20 trials Preferred reinforcer delivered for 10 s on a fixed 1:1 schedule Tutor/parent placed arm across child's arms to prevent them from knocking spoon	✓	✓	✓	**





Appendix I *Continued*

Study	Goal (therapy agent)	Treatment design	Antecedent	Definition	Consequence	Author definition	Positive	Negative	Reinforcement	Punishment
Milnes (2011)	Replicate the procedure used by Gentry and Luiselli (2008). Antecedent manipulation + CR (parent)	Random chance game to determine session volume → CR + escape after randomly determined volume of food eaten	Mystery motivator spinner Verbal prompt	Child spun chart with different numbers to decide number of bites 'You have spun number 2. That means you can eat 2 bites from this section, 2 bites from this section and 2 bites from this section. Once you finish, you can eat whatever you like, or leave the table and play with X' Child touching food to lips and tasting rather than eating were used as a progression towards accepting bites in some cases	CR (object)  Escape  CR (verbal)	Rewards provided contingent on completing task set by the mystery motivator spinner  Child had option to leave table after finishing required bites Parents praised following first bite of food accepted 'Good job, you have N more bites to eat and then you get X' If child did not meet target number of bites, they had to remain at table for 15 min with praise and reward withheld Slow increase in volumes presented for one child	✓	✓	✓	✓
Najdowski <i>et al.</i> (2003)	Compare prompt + CR vs. prompt + NRS + CR (parent)	Prompt + CR (BL) vs. Prompt + NRS + CR + thinning Session concluded when required volume consumed (minimum 1 bite) or after 30 min	3-step prompting procedure NRS	1. Instruction to self-feed 2. Modelling how to take a bite 3. Physical prompt Food held at mouth until accepted or 30 min elapsed	CR (verbal and food)  Thinning reinforcement	Praise provided Plate full of preferred foods provided Total number of bites required increased as intervention became more successful (increased by 50% every 3 successful sessions)	✓	✓	✓	✓
Najdowski <i>et al.</i> (2010)	Compare prompt + CR vs. NRS + CR (parent) Vs NRS + CR + thinning Session concluded when required volume consumed (minimum 1 bite) or after 30 min	Prompt + CR + Escape (BL) Vs NRS + CR + thinning Session concluded when required volume consumed (minimum 1 bite) or after 30 min	Hunger manipulation 3 step prompting procedure (BL) NRS	No food permitted in 3–4 h prior to sessions 1. Instruction to self-feed 2. Modelling how to take a bite 3. Physical prompt Food held at lips until child accepted	CR (food)  Thinning reinforcement  Ignoring  Re-presentation Escape (BL)  CR (verbal)	Plate of highly preferred foods provided Number of bites required before reinforcement increased systematically Attention withdrawn in response to undesirable behaviours Expelled foods re-presented Escape provided contingent on undesirable behaviours Praise provided if bites accepted	✓	✓	✓	✓
Najdowski <i>et al.</i> (2012)	Evaluate prompt + CR + texture grading +/- simultaneous presentation (therapist)	Prompt + CR + texture grading +/- simultaneous presentation after 20 bite presentations	Verbal prompt Tactile prompt Grading (texture) Simultaneous presentation	'Open' Spoon presented to bottom lip Texture of foods slowly increased to chopped consistency Preferred texture presented at same time as non-preferred texture	CR (object)  Escape	Access to highly preferred video provided for 15 s If bite not accepted in 5 s, it was removed for 25 s and then new bite presented	✓	✓	✓	✓
Patel <i>et al.</i> (2007)	Evaluate high probability-low probability instructional sequence (therapist)	High-probability sequences (shaping) Session concluded after 5 low-probability bites accepted	High-probability request  Low-probability request	'Take a bite' from an empty spoon  'Take a bite' of spoon with food 3:1 ratio for high probability to low probability requests	CR (verbal + touch)	Verbal praise and light physical touch delivered if request complied with on a fixed 1:1 schedule	✓	✓	✓	✓

Paul et al. (2007)	Evaluate intervention combining repeated taste exposure and escape prevention (therapist)	Taste meals (single presentation with the expectation to take a bite, with escape as a reward) and probe meals (10 min meal presentation with no expectation to eat) conducted. Taste sessions concluded after 1 bite. Probe sessions concluded after 10 min.	Verbal prompt Grading (volume)	'When you take your bite, you can go play' Size of bite slowly increased contingent on multiple acceptances (minimum 'pea-sized' bite)	Escape (taste meals) Planned ignoring CR (verbal) Escape (probe meals)	Child was allowed to leave contingent on acceptance of bite on a fixed 1:1 schedule Inappropriate behaviours ignored Praise provided if bite was taken on a fixed 1:1 schedule If child did not taste one or more goal foods, they were allowed a 5 min break before taste sessions began again Child was allowed to leave contingent on acceptance of bite Inappropriate behaviours ignored Praise provided if bite taken	✓ ✓ ✓ ✓	✓ ✓ ✓ ✓	✓ ✓ ✓ ✓
Pizzo et al. (2009)	Replicate findings of Paul et al. in a shorter time frame (therapist)	Taste meals (single presentation with the expectation to take a bite, with escape as a reward) and probe meals (10 min meal presentation with no expectation to eat) conducted. Taste sessions concluded after 1 bite. Probe sessions concluded after 10 min. Multiple taste sessions/day (single bite followed by escape) Probe meal after 10 taste sessions (foods presented without pressure to eat) Taste sessions concluded after 1 bite. Probe sessions concluded after 10 min.	Verbal prompt Grading (volume)	Child told they would be able to leave when bite accepted on multiple acceptances (minimum 'pea-sized' bite)	Escape (taste meals) Planned ignoring CR (verbal)	Child was allowed to leave contingent on acceptance of bite Inappropriate behaviours ignored Praise provided if bite taken	✓ ✓ ✓	✓ ✓ ✓	✓ ✓ ✓
Seiverling et al. (2012)	Evaluate repeated taste exposure and volume grading (parent)	Multiple taste sessions/day (single bite followed by escape) Probe meal after 10 taste sessions (foods presented without pressure to eat) Taste sessions concluded after 1 bite. Probe sessions concluded after 10 min.	Hunger manipulation Grading (volume)	Refrain from providing non-target foods for 2 h before/after daily taste sessions Parent was expected to increase volume on spoon if child accepted bite within 30 s (minimum 'pea-sized' bite)	Escape Ignoring Re-presentation CR (verbal)	Child could leave for 3 min after taste accepted Parent instructed to ignore disruption Parent re-presented expelled bites Praise for acceptance of bites provided	✓ ✓ ✓ ✓	✓ ✓ ✓ ✓	✓ ✓ ✓ ✓
Sharp and Jacquess (2009)	Evaluate antecedent-based treatment changes on volume and texture (therapist)	Prior to reported treatment, the child had undergone Rx using NRS + NCR to teach bite acceptance. Applied same operant techniques as above, but manipulated antecedent. Session concluded after pre-determined volume consumed (unclear?). Non-removal of the meal + escape when child finished meal, or if it became time for another unavoidable activity	NRS Grading (volume) Grading (texture)	Bite positioned at lips until child opened mouth; therapist followed child's mouth with the spoon in response to head-turns. Increased bite size from '2-pea' size, to half-level, level and rounded spoon. Increased texture from pureed, to wet ground, to ground, to chopped.	Blocking Re-presentation NCR	Therapist physically stopped disruptive behaviours Expelled bites represented (for volume grading only) Access to highly preferred leisure items provided throughout treatment	✓ ✓ ✓	✓ ✓ ✓	✓ ✓ *
Tarbox et al. (2010)	Evaluate non-removal of the meal (parent)	Non-removal of the meal + escape when child finished meal, or if it became time for another unavoidable activity	Verbal prompt Non-removal of the meal	Scripted verbal prompt provided. Meal was not removed until completed, or until another activity which could not be rescheduled occurred	Physical prompt Escape Re-presentation	If child attempted to leave table, parent physically returned him to table If meal completed, child allowed to leave table If meal was not completed before another scheduled activity, the same meal was re-presented at the next scheduled session	✓ ✓ ✓	✓ ✓ ✓	✓ ✓ ✓

**Appendix I** *Continued*

Study	Goal (therapy agent)	Treatment design	Antecedent	Definition	Consequence	Author definition	Positive	Negative	Reinforcement	Punishment
Validmarsdotir et al. (2010)	To replicate the findings of Najdowski et al. (2003) (Therapist/teachers/parent)	Prompt + CR/Escape + NRS/ Re-presentation Session concluded when pre-determined volume consumed (minimum 1 bite), or after 30 min	Verbal prompt	Prompt to take a bite provided every 30 s Feeder told child number of bites required to receive reinforcement	CR (verbal) CR (object)	Provided immediately after accepting each bite on a 1:1 fixed schedule Provided after each bite accepted, and thinned to a token system as success achieved	✓	✓	✓	✓
Volkert et al. (2011)	Evaluate redistribution and swallow facilitation techniques to decrease food packing (therapist)	Prior to this study, a programme using NRS was administered to increase acceptance. Packing emerged as a response to increased texture. NRS + flipped spoon + CR Session concluded after 25 bite presentations	NRS	Bite presented at child's lips until accepted	CR (verbal) Flipped spoon	Number of bites required to receive reinforcement increased in a systematic manner as success achieved If child did not take bite, fork was held close to mouth until bite consumed If child spat bite out, feeder presented new bite Praise provided if bite accepted, and if mouth cleared on a 1:1 fixed schedule If food remained in mouth, spoon inserted and food redistributed to centre of tongue. Firm pressure applied while spoon was dragged anteriorly. Inappropriate behaviours were ignored Expelled bites were re-presented	✓	✓	✓	✓
Wildert et al. (2005)	Compare NCR (without NRS) + CR+ escape vs NCR + NRS + CR + escape (therapist)	NRS + CR + Escape vs. NRS + NCR + CR + Escape Session concluded after 5 min	NRS	Therapist presented bite of food every 30 s Bite remained at lips for 30 s if it was not accepted, and the child did not engage in self-injury	CR (verbal) Escape NCR	Brief praise delivered if child accepted bite Contingent on self-injury, spoon removed and therapist moved away for 15 s Child had continuous access to children's video	✓	✓	✓	**
Wood et al. (2009)	Evaluate gradual introduction of new foods with CR (therapist)	Prompting + shaping + CR Session concluded after 10 bite presentations	3 stage prompting Shaping (preferred → non-preferred)	1. 'Take a bite' 2. Physical assistance (hand over hand to get spoon) 3. Physical assistance (hand over hand to put spoon to mouth) Moved through four categories of food preferences	CR (verbal) Escape Bite size reduction Touch tongue	Praise provided if bite accepted Spoon returned to plate if bite not accepted Smaller bite presented if child refused Child asked to touch tongue to food if refusing to accept food	✓	✓	✓	✓

NRS = non-removal of the spoon; CR = contingent reinforcement; NCR = non-contingent reinforcement; BL = baseline condition; NPF = non-preferred food.

Intervention features as defined by Marshall *et al.*

Intervention feature	Definition
Antecedent	Stimulus which elicits a response from the child (e.g. presenting the spoon)
Response	Child's behaviour as a result of antecedent, e.g. accepting bite vs. screaming (often measured as the dependent variable)
Consequence	Adult response to child behaviour. May be either reinforcement or punishment
Reinforcement	Consequence applied if the desire is for the behaviour to occur again
Punishment	Consequence applied if the desire is for the behaviour not to occur again
Non-removal of the spoon	Spoon remains at the child's lips until they accept the bite. This is a form of escape extinction. It is designed to be an antecedent, but could also be a consequence (i.e. negative reinforcement – the spoon is removed after the child accepts the bite)
Thinning reinforcement	The expected response for reinforcement increases (e.g. previously 1:1 reinforcement increases to 2:1)
*Non-contingent reinforcement	Child has access to preferred objects during session which are not provided contingent on behaviour (i.e. distraction). Although this is reinforcing for the child, it is not strictly defined as a reinforcer, given it is not contingent on a behaviour occurring before reinforcement is provided
**Escape as a negative punishment	In some cases, escape was provided in response to refusal or self-injury. It was assumed that provision of escape in these situations was designed as a 'time-out' from the feeding situation
	In some cases, escape was provided in a non-contingent fashion (e.g. every 30 s regardless of child behaviour).

**Appendix II**

Extended table of quality review responses

Study	Participants/settings			Dependent variable				Independent variable			
	Participants described with sufficient detail	Process for selecting participants replicable	Critical features of physical setting described	DV(s) described with operational precision	DV measured in a way that generates quantifiable index	Measurement of DV valid and replicable	DV is measured repeatedly	IOA >80% across all DV	IV is replicable	IV systematically manipulated and in control of experimenter	Fidelity of implementation described
Ahearn et al. (2001)	✓			✓	✓	✓	✓	✓	✓		✓
Allison et al. (2012)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Anderson and McMillan (2001)	✓	✓		✓	✓	✓	✓	✓	✓		✓
Bui et al. (2013)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Gale et al. (2011)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Gentry and Luiselli (2008)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Levin and Carr (2001)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
McCarthy et al. (2005)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Meier et al. (2012)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Milnes (2011)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Najdowski et al. (2003)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Najdowski et al. (2010)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Najdowski et al. (2012)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Patel et al. (2007)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Paul et al. (2007)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Pizzo et al. (2009)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Seiverling et al. (2012)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Sharp and Jacquess (2009)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Tarbox et al. (2010)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Valdimarsdottir et al. (2010)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Volkert et al. (2011)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Wood et al. (2009)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Wilder et al. (2005)	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓

Study	Baseline		Internal validity		External validity		Social validity		IV implemented over extended time periods, by typical intervention agents, or in typical social contexts	Total (/21)
	BL present	BL replicable	Experimental effect demonstrated at 3 time points	Design controls for threats to internal validity	Results demonstrate experimental control	Effects replicated across participants, settings or materials	DV socially important	Magnitude of change of DV socially important		
Ahearn et al. (2001)	✓	✓			✓	✓	✓	N/D	✓	14
Allison et al. (2012)	✓	✓		✓	✓	✓	✓	N/D	✓	15
Anderson and McMillan (2001)	✓	✓		✓	✓	✓	✓	N/D	✓	12
Bui et al. (2013)	✓	✓	✓	✓	✓	✓	✓	N/D	✓	14
Gale et al. (2011)	✓	✓		✓	✓	✓	✓	N/D	✓	18
Gentry and Luiselli (2008)	✓	✓		✓	✓	✓	✓	N/D	✓	14
Levin and Carr (2001)	✓	✓		✓	✓	✓	✓	N/D	✓	16
McCarthy et al. (2005)	✓	✓		✓	✓	✓	✓	N/D	✓	18
Meier et al. (2012)	✓	✓		✓	✓	✓	✓	N/D	✓	15
Milnes (2011)	✓	✓		✓	✓	✓	✓	N/D	✓	14
Najdowski et al. (2003)	✓	✓		✓	✓	✓	✓	N/D	✓	15
Najdowski et al. (2010)	✓	✓		✓	✓	✓	✓	N/D	✓	15
Najdowski et al. (2012)	✓	✓		✓	✓	✓	✓	N/D	✓	14
Patel et al. (2007)	✓	✓		✓	✓	✓	✓	N/D	✓	15
Paul et al. (2007)	✓	✓		✓	✓	✓	✓	N/D	✓	7
Pizzo et al. (2009)	✓	✓		✓	✓	✓	✓	N/D	✓	13
Seiverling et al. (2012)	✓	✓		✓	✓	✓	✓	N/D	✓	13
Sharp and Jacquess (2009)	✓	✓		✓	✓	✓	✓	N/D	✓	18
Tarbox et al. (2010)	✓	✓		✓	✓	✓	✓	N/D	✓	14
Valdimarsdottir et al. (2010)	✓	✓		✓	✓	✓	✓	N/D	✓	16
Volkert et al. (2011)	✓	✓		✓	✓	✓	✓	N/D	✓	16
Wood et al. (2009)	✓	✓		✓	✓	✓	✓	N/D	✓	14
Wilder et al. (2005)	✓	✓		✓	✓	✓	✓	N/D	✓	13

DV, dependent variable; IV, independent variable; IOA, inter-observer agreement; BL, baseline; N/D, not able to be determined.

## Appendix III

Raw data for Improvement Rate Difference (IRD) calculations

Study	DV	Increase desirable behaviour	Decrease undesirable behaviour	Phases	Phase number	Improved baseline	Total baseline	Improved treatment	Total treatment	IRD	Overall IRD	IRD lower 95% confidence interval	IRD upper 95% confidence interval	Notes
Ahearn <i>et al.</i> (2001)	% bites accepted	✓		3	1	5	10	7	14	0				% expulsions not presented in visual analysis
					2	1	3	6	14	0.10				
Allison <i>et al.</i> (2012)	% occurrence disruption	✓		3	3	1	3	20	23	0.54	0.17	0.12	0.45	
					1	1	10	7	14	0.4				
					2	3.5	4	3.5	15	-0.64				
					3	1	3	20	23	0.54				
					1	3	10	6	14	0.13				
					2	3.5	4	2.5	15	-0.71				
Anderson and McMillan (2001)	% occurrence negative vocalization	✓		3	3	3.5	4	13.5	24	-0.31	-0.12	-0.28	0.04	
					1	0.5	4	16.5	17	0.85				
					2	0.5	4	16.5	17	0.85				
					3	0.5	4	16.5	17	0.85				
					1	1.5	4	16.5	17	0.60				
					2	0.5	4	16.5	17	0.85				
Bui <i>et al.</i> (2013)	% bites accepted (breakfast) (lunch) (dinner)	✓		3	3	0.5	4	16.5	17	0.85	0.85	0.65	1.04	
					1	0.5	4	15.5	16	0.89				
					2	0.5	4	15.5	16	0.84				
					3	0.5	4	15.5	16	0.81				
					1	5	6	14	15	0.1				
					2	2	3	1	15	-0.6				
Gale <i>et al.</i> (2011)	% bites expelled	✓		3	3	2.5	4	0.5	9	-0.57				
					1	4	5	14	16	0.08				
					2	3.5	4	14.5	17	-0.02				
					3	3.5	4	6.5	9	-0.15				
					1	2	5	15	16	0.54				
					2	1	3	14	16	0.54				
Gentry and Luiselli (2008)	% bites with self-injurious behaviour	✓		3	3	0.5	4	8.5	9	0.82	0.13	-0.003	0.27	
					1	0.5	4	6.5	7	0.80				
					1	0.5	5	8.5	9	0.84				
					1	0.5	6	9.5	10	0.87				
					1	0.5	10	57.5	59	0.98				
					1	0.5	15	54.5	60	0.92				
Gentry and Luiselli (2008)	% bites accepted (breakfast) (lunch) (dinner)	✓		3	3	0.5	19	28.5	51	0.56	0.85	0.67	1.02	Excluded sessions 1 and 8 for John due to author instructions
					1	0.5	19	28.5	51	0.91				
					1	0.5	10	56.5	59	0.91				
					1	0.5	15	54.5	60	0.8				
					1	0.5	19	35.5	51	0.67				
					1	0.5	6	47.5	53	0.81				



Appendix III *Continued*

Study	DV	Increase desirable behaviour	Decrease undesirable behaviour	Phases	Phase number	Improved baseline	Total baseline	Improved treatment	Total treatment	IRD	Overall IRD	IRD lower 95% confidence interval	IRD upper 95% confidence interval	Notes
Levin and Carr (2001)	Grams of food consumed (Luis)	✓		1	1	0.5	12	21.5	25	0.82	0.82	0.64	1.00	
	Frequency of problem behaviour		✓	1	1	10.5	12	24.5	25	0.11	0.11	-0.09	0.30	
Meier <i>et al.</i> (2012)	% acceptance for plums	✓		3	1	0.5	4	3.5	4	0.75	0.75			
	% acceptance for raspberries	✓		1	2	1	6	2	3	0.5	0.5			
	% acceptance for eggplant	✓		1	3	2	6	11	12	0.58	0.58			
	% acceptance for # bites consumed (Brian)	✓		1	1	0.5	5	10.5	11	0.85	0.85			
Milnes (2011)	% acceptance for # bites consumed (Lawrence)	✓		1	1	0.5	6	21.5	24	0.81	0.76	0.61	0.92	
	% intervals with verbal refusal (Brian)	✓		1	1	2	13	55	61	0.75	0.75			
	% intervals with verbal refusal (Lawrence)	✓		1	1	2	7	35	79	0.16	0.59	0.41	0.77	
	% intervals out of seat (Brian)	✓		1	1	7	13	78	84	0.39	0.39			
	% intervals out of seat (Lawrence)	✓		1	1	6	7	80	82	0.12	0.12			
McCartney <i>et al.</i> (2005)	Frequency of bites (Matt)	✓		1	1	11	13	79	83	0.11	0.11			
	Frequency of bites (Kurt)	✓		1	1	6	7	87	90	0.11	0.16	0.04	0.29	
	Frequency of expulsions (Matt)	✓		1	1	2.5	5	57.5	58	0.49	0.49			
	Intervals with interruptions (Matt)	✓		1	1	0.5	4	105.5	106	0.87	0.74	0.48	1.00	
	Frequency of expulsions (Kurt)	✓		1	1	3.5	4	105.5	106	0.12	0.12			
	Intervals with interruptions (Kurt)	✓		1	1	1.5	7	105.5	106	0.78	0.48	0.32	0.64	
Najdowski <i>et al.</i> (2003)	# bites accepted (home)	✓		1	1	0.5	6	57.5	63	0.83	0.83			
	# bites accepted (restaurant)	✓		1	1	0.5	3	16.5	17	0.80	0.82	0.62	1.03	
Najdowski <i>et al.</i> (2010)	% bites swallowed (Annabelle)	✓		1	1	0.5	3	34.5	39	0.72	0.72			
	% bites swallowed (Collin)	✓		1	1	0.5	5	31.5	37	0.75	0.74	0.50	0.98	
Najdowski <i>et al.</i> (2012)	% mouth clean (food set 1)	✓		3	1	1.5	5	4.5	5	0.6	0.6			
	% mouth clean (food set 2)	✓		3	2	1.5	4	42.5	43	0.53	0.61			
	% bite acceptance	✓		3	1	2.5	5	6.5	7	0.43	0.41	0.22	0.61	
Patel <i>et al.</i> (2007)	% bite acceptance	✓		3	2	3.5	5	6.5	7	0.23	0.19	0.22	0.61	
		✓		3	3	0.5	8	6.5	7	0.87	0.86	0.71	1.01	
		✓		3	2	0.5	7	6.5	7	0.86	0.86	0.71	1.01	

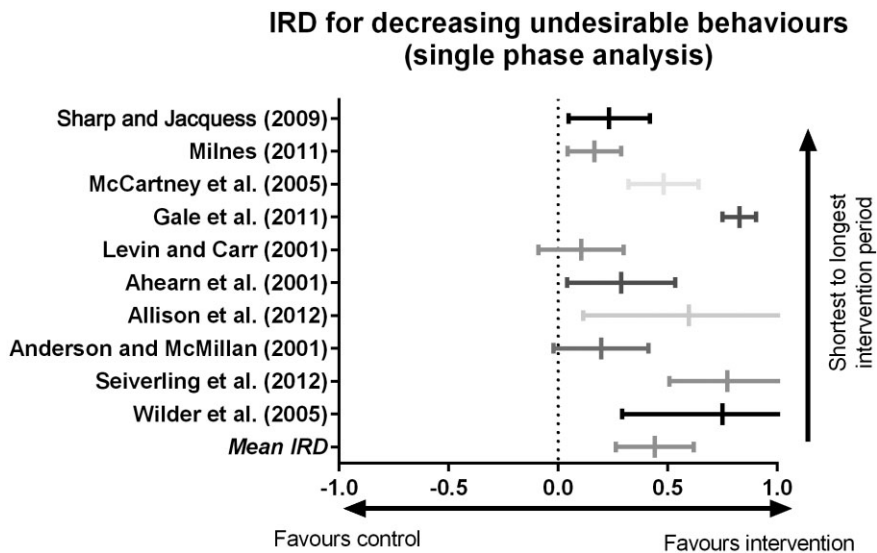
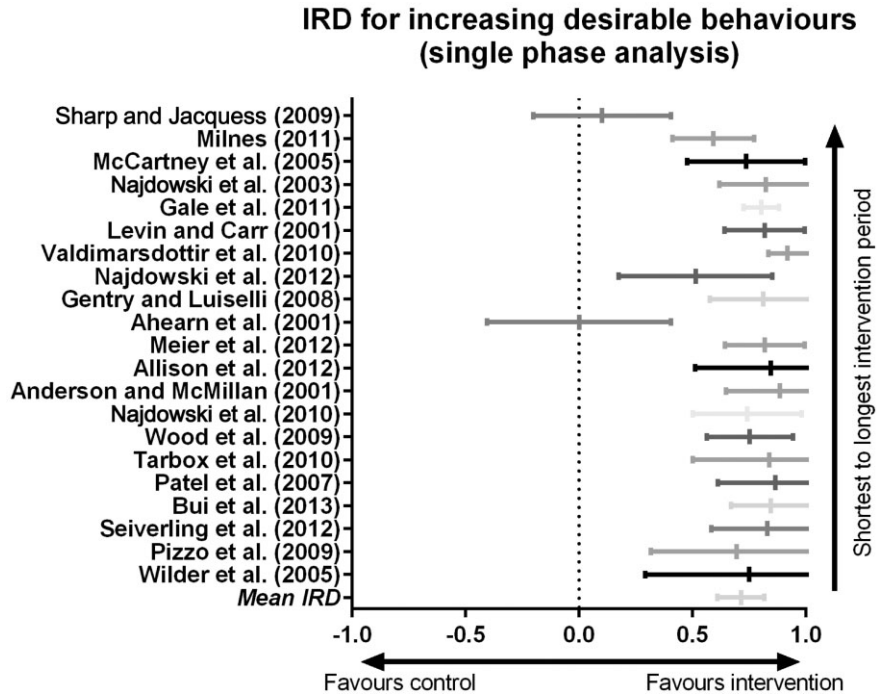
Pizzo <i>et al.</i> (2009)	✓	# bites accepted (Sam)	1	1	0.5	9	4.5	6	0.69	0.69	0.32	1.07
Seiverling <i>et al.</i> (2012)	✓	# bites accepted (Tommy)	1	1	0.5	3	9.5	10	0.78			
	✓	# bites accepted (Noah)	1	1	0.5	5	9.5	10	0.85	0.83	0.58	1.08
	✓	Proportion accepted bites with disruptive behaviours (Tommy)	1	1	0.5	3	10.5	11	0.79			
	✓	Proportion accepted bites with disruptive behaviours (Noah)	1	1	0.5	5	9.5	11	0.76	0.77	0.51	1.04
Sharp and Jacques (2009)	✓	% mouth clean (volume)	1	1	0.5	3	27.5	28	0.15			
	✓	% mouth clean (texture)	1	1	0.5	3	35.5	40	0.05	0.10	-0.20	0.41
	✓	Difficult behaviours per bite (volume)	1	1	0.5	3	21.5	28	0.60			
	✓	Difficult behaviours per bite (texture)	1	1	2.5	3	13.5	40	-0.50			
	✓	Expulsions per bite (volume)	1	1	2.5	3	27.5	28	0.15			
	✓	Expulsions per bite (texture)	1	1	2.5	3	33.5	40	0.00			
	✓	Gags per bite (volume)	1	1	0.5	3	25.5	28	0.74			
	✓	Gags per bite (texture)	1	1	1.5	3	39.5	40	0.49	0.23	0.05	0.42
Tarbox <i>et al.</i> (2010)	✓	% meal consumed	3	1	0.5	4	13.5	14	0.84			
	✓	# bites consumed (preschool)	1	1	0.5	2	13.5	15	0.71			
	✓	# bites consumed (home)	1	1	0.5	7	34.5	38	0.65	0.78	0.51	1.05
	✓	% bite acceptance	1	1	0.5	27	29.5	31	0.93	0.92	0.83	1.00
Wildner <i>et al.</i> (2005)	✓	% intervals with self-injurious behaviour	3	1	0.5	4	3.5	4	0.75			
	✓	% acceptance Cat 1	1	1	0.5	4	3.5	4	0.75			
	✓	% acceptance Cat 2	1	1	0.5	4	3.5	4	0.75	0.75	0.49	1.01
	✓	% acceptance Cat 3	1	1	0.5	4	3.5	4	0.75			
	✓	% acceptance Cat 4	1	1	0.5	4	3.5	4	0.75	0.75	0.49	1.01
Wood <i>et al.</i> (2009)	✓	% acceptance Cat 1	1	1	1.5	5	32.5	33	0.68			
	✓	% acceptance Cat 2	1	1	0.5	4	14.5	16	0.78			
	✓	% acceptance Cat 3	1	1	0.5	4	6.5	7	0.80			
	✓	% acceptance Cat 4	1	1	0.5	4	8.5	10	0.73	0.75	0.56	0.94

DV, dependent variable; IRD, Improvement Rate Difference.  
Please note that cells containing zero (e.g. no events in one group) caused problems with calculating standard errors. As a result we added 0.5 to each cell of the grid for any such study (Higgins & Green 2011).  
Higgins, J. P. T. & Green, S. (2011) *Cochrane Handbook for Systematic Reviews of Interventions*. The Cochrane Collaboration.

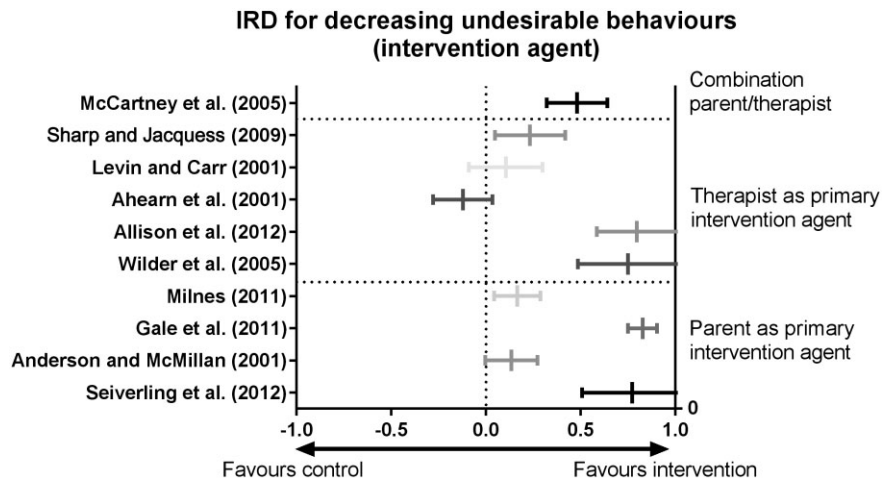
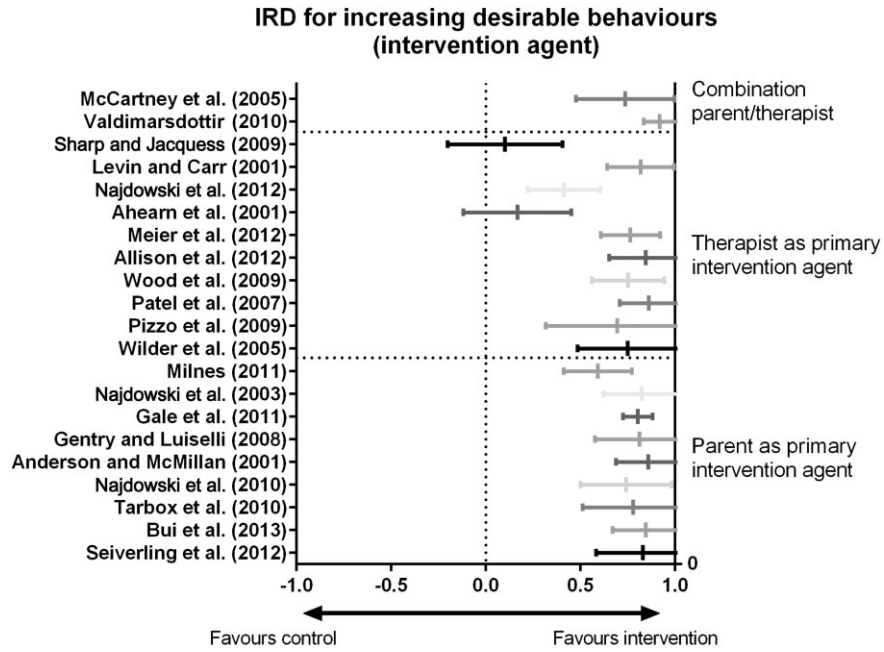
## Appendix IV

### Improvement Rate Difference (IRD) sensitivity analyses

#### 1. IRD for single phase analysis by intervention duration



2. IRD for parent vs. therapist as agent of change



3. IRD comparing intensity of therapy delivery

