Systematic Review of Psychological Interventions for Pediatric Feeding Problems

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Objective To conduct a systematic review of the research evaluating the effect of psychological interventions for pediatric feeding problems. Methods A search was conducted to identify studies using psychological interventions for pediatric feeding problems published between 1998 and 2013. Randomized controlled trials (RCTs) and nonrandomized studies that examined aggregated outcome data were included. Primary outcomes were child mealtime behavior, nutritional status, and caregiver stress. A risk of bias assessment was conducted and the quality of the evidence rated using Grading of Recommendations Assessment, Development, and Evaluation methodology. Results 13 studies were identified and a narrative synthesis framework was used to report findings. Conclusions The preponderance of evidence suggests positive effects of psychological intervention for the treatment of feeding problems. However, limited data and the paucity of studies using RCT methodologies limit conclusions that can be drawn regarding the efficacy of these interventions. Future studies using more rigorous research methods are needed to enhance understanding of these interventions.

Key words evidence-based practice; gastroenterology; nutrition; systematic review.

Background
Feeding problems are identified if a child’s eating behavior has an adverse effect on health or psychosocial functioning (Bryant-Waugh, Markham, Kreipe, & Walsh, 2010; Kedesdy & Budd, 1998). The behavioral manifestations of feeding problems, and ultimately the targets of psychological intervention, include food refusal, food selectivity, or other disruptive behaviors incompatible with eating. Feeding problems are observed most commonly in early childhood but can persist into school age and adolescence if unaddressed (Babbitt et al., 1994; Dahl & Sundelin, 1992).

Feeding problems are reported in approximately 25–45% of the general population (Bentovim, 1970), occur in up to 80% of children with developmental disabilities (Manikam & Perman, 2000), and are estimated to occur in 40–70% of children with chronic medical conditions (Davis, Bruce, Cocjin, Mousa, & Hyman, 2010; Douglas & Bryon, 1996; Thommessen, Heiberg, Kase, Larsen, & Riis, 1991). Feeding disorders, characterized by chronic feeding problems, which jeopardize long-term growth and nutrition, are estimated to affect 3–5% of children, making feeding disorders one of the most common conditions presenting as a concern to pediatricians (Satter, 1990).

Etiological contributors to feeding problems include medical, anatomical, developmental, temperamental, social, and environmental factors. Difficulties in any of these areas can interrupt or delay typical feeding development, resulting in poor nutrition. Subsequently, poor nutrition can impact weight gain, linear growth, and other functional health outcomes as well as cognitive development and emotional regulation (Silverman & Tarbell, 2009). Caregiver stress is also commonly associated with childhood feeding problems (Garro, Thurman, Kerwin, & Ducette, 2005). Finally, feeding
problems represent a significant financial burden to families in terms of direct medical expense and lost work for adult caregivers (Williams, Riegel, Gibbons, & Field, 2007). Overall, the relative risks associated with untreated feeding problems are pronounced, supporting the need for early intervention.

The complexity of feeding problems warrants interdisciplinary intervention. Physicians, dietitians, speech and language pathologists, and occupational therapists provide treatment focused on medical management and skill-deficit remediation. However, once medical and mechanical factors have been identified and ameliorated, challenging mealtime behaviors often persist (Babbitt et al., 1994). This maintenance of maladaptive mealtime behaviors by environmental factors is often addressed by the use of psychological and behavioral interventions that have been well described in prior review articles (Kedesdy & Budd, 1998; Kerwin, 1999; Linscheid, 2006; Sharp, Jaquess, Morton, & Herzinger, 2010; Vaz & Piazza, 2011). Behavioral interventions manipulate antecedent situations and environmental contingencies with goals of changing maladaptive mealtime behavior and subsequently improving nutritional intake, weight gain, and linear growth. Antecedent and contingency management strategies typically include appetite manipulation, differential attention, escape prevention, and systematic desensitization (Williams, Field, & Seiverling, 2010).

Intervention environments described in the literature can broadly be classified as outpatient, day treatment, or inpatient. Traditional outpatient settings are generally more accessible and less expensive than other models of care. Intensive or day treatment models allow for greater frequency of contact and tighter control over the feeding environment and increase opportunities for coordinated care across disciplines. Inpatient hospitalization allows for the greatest environmental control and the closest medical oversight, making these settings ideal for the treatment of the most difficult cases or for cases that require close medical collaboration.

Five prior reviews summarizing the evidence for psychological intervention in the treatment of pediatric feeding problems have been published, each evaluating the literature in a different manner, yet each supporting the use of behavioral or psychological intervention. The first systematic review of the literature was conducted by Kerwin in 1999. Kerwin reviewed psychosocial and behavioral intervention studies published between 1970 and 1997 using criteria created by the Task Force on the Promotion and Dissemination of Psychological Procedure (1995). Ledford and Gast conducted a subsequent review in 2006 and identified nine intervention studies published between 1994 and 2000 focusing on the psychological treatment of feeding problems in children with autism spectrum disorders. Williams and colleagues (2010) conducted a third review of studies published between 1979 and 2008 and identified 38 intervention studies that included at least one behavioral component. Davis et al. (2010) conducted a review separately examining interventions for children who fed orally and children who presented with dependence on gastrostomy tube feedings. Further, Davis and colleagues elaborated on the methodology of primary studies, noting that the majority of studies used chart review techniques with only one study of outpatient intervention conducted as a randomized controlled trial (RCT) (Benoit, Want, & Zlotkin, 2000). Finally, to further synthesize treatment outcomes, Sharp and colleagues (2010) conducted a meta-analytic review of 48 psychological intervention studies published between 1970 and 2010 that used single-case designs with an element of experimental control.

The conclusion of these review studies was that behaviorally based interventions are the most well documented and empirically supported interventions for pediatric feeding problems. However, there are methodological limitations impacting conclusions that can be drawn from both primary and review studies. For example, the evidence base is primarily composed of observational studies, with most using single-subject design. Those studies not employing single-subject design lack adequate power owing to small sample sizes. Finally, many primary studies and review studies focus solely on outcomes related to behavior change with minimal attention devoted to outcomes related to nutrition, physical health, psychological well-being, and family functioning.

The purpose of this review is to summarize the findings of studies that examine psychological interventions that target maladaptive mealtime behavior, aim to improve oral nutrition, and address psychosocial variables associated with pediatric feeding problems. This review uses a narrative synthesis framework to summarize the findings of primary studies and uses Grading of Recommendations Assessment, Development, and Evaluation (GRADE) methodology to estimate the overall quality of the existing evidence. This review also identifies methodological limitations of current studies and serves to set an agenda for future and more controlled research.

Method
Criteria for Considering Studies for This Review
Types of Studies
Studies of interventions that were primarily psychological in nature targeting pediatric feeding problems that were
published in a peer-reviewed journal were considered for inclusion. Given that the efficacy of behavioral intervention for the treatment of pediatric feeding problems has not been adequately examined in RCTs, both RCTs and nonrandomized studies (NRS) were included in this review. NRS were included following guidelines of the Cochrane Collaboration that a systematic review should include the best available study designs with the least risk of bias (Reeves, Deeks, Higgins, & Wells, 2011). As there are great concerns regarding threats to validity in NRS, specific study design features of included studies were carefully examined to clearly identify potential sources of bias.

Specifically, RCTs that compared psychological intervention with attention control, waiting list control, or other active treatment (including nutritional or medical) with at least 10 participants in each of the treatment and control arms were considered for inclusion. NRS that examined outcomes within a group of participants over time were also included if the outcome data for participants was aggregated, rather than presented as a case series.

**Types of Participants**
To be considered in the review, the primary presenting problem for participants was a pediatric feeding problem in children birth through 18 years of age. Exclusion criteria were animal studies or participants presenting with a primary diagnosis of anorexia nervosa, bulimia nervosa, or eating disorder not otherwise specified.

**Types of Interventions**
Included studies evaluated interventions designed to alter child and/or caregiver behavior and were individual in nature, conducted in a group, or included a parent training component. Interventions considered for review were predominantly psychological in nature, employing components such as behavioral therapy, parent training, play therapy, or family therapy. Given the complex interplay among etiological factors contributing to feeding problems and that behavioral intervention often includes antecedent manipulation, the review also included studies that used nutritional manipulation (e.g., tube weaning), as this can potentially influence a child’s eating behavior.

**Types of Outcome Measures**
The primary goal of behavioral intervention for the treatment of pediatric feeding problems is to alter mealtime behavior that interferes with adequate nutritional intake and adversely impacts parent–child interaction at mealtimes. As such, child mealtime behavior was a primary outcome domain of interest, evaluated via directly observed behavior and parent report of mealtime behavior. Nutritional status (e.g., dependence on supplemental nutrition, caloric intake) was also considered. Finally, measures of psychosocial variables associated with pediatric feeding problems (e.g., caregiver stress) were included.

**Search Methods for Identification of Studies**
The search strategy included a review of the following electronic databases, searching dates between January 1998 and April 2013: MEDLINE, The Cochrane Central Register of Controlled Trials (CENTRAL), and PsycINFO. January 1998 was selected as the earliest date for this review, as it was the end-date for studies included in the comprehensive review conducted by Kerwin in 1999. Searches of the electronic databases were conducted using the keyword function in each database. Search terms related to pediatric feeding problems (feeding disorder, selective eating, dysphagia, food aversion, food refusal, neophobia, food phobia, sensory food aversion, feeding problem, pediatric feeding problem, failure to thrive, faddy eating, under-nutrition, and infantile anorexia) were systematically paired with search terms related to psychological intervention (psychological intervention, psychological treatment, behavioral intervention, and behavioral treatment). Prior meta-analyses and reference lists from identified studies were also reviewed.

**Data Collection and Analysis**

**Selection of Studies**
Two research assistants identified studies through database searching as described above, and duplicates were removed using reference management software (Endnote X7). Another research assistant screened titles and abstracts of studies from the original database searches for inclusion in the review based on predetermined inclusion and exclusion criteria reported above, reviewing full articles if inclusion could not be determined from title and abstract alone. Reliability of decision making was evaluated by the authors for 30% of the included studies and 30% of the excluded studies. Cohen’s kappa was .93 (p = .000) indicating strong inter-rater agreement. In the case of discrepancies, the final decision regarding inclusion was made by the lead author.

**Data Extraction and Management**
The authors conducted data extraction and bias assessment independently for all included studies using a data extraction form designed for this review. The form included prespecified study components such as details of the reference, study design, participants and demographic/descriptive data, diagnosis, type of intervention, target of
intervention, treatment components, treatment setting, outcome measures, statistical analyses, and conclusions of the authors based on statistical analyses. The authors evaluated reliability of the data extraction for all of the included studies and resolved discrepancies by consulting the original articles.

Assessment of Risk of Bias in Included Studies
As NRS were a major focus of this review, the authors carefully examined the unique risks of bias most commonly associated with NRS (selection, performance, detection, attrition, and reporting biases) using multiple methods of bias assessment as recommended by the Cochrane Collaboration (Reeves et al., 2011). First, the specific design features of each study (e.g., prospective generation of hypotheses) were identified and associated risks of bias reported. Secondly, the Cochrane Risk of Bias Tool was adapted to evaluate bias most commonly associated with NRS (Higgins, Altman, & Sterne, 2011). The authors evaluated risk of bias independently for each study. Disagreements were discussed between the authors and original articles consulted to resolve discrepancies.

Quality of the Evidence
The authors used the GRADE system to evaluate the overall quality of the evidence supporting the use of psychological and behavioral interventions for the treatment of pediatric feeding problems. GRADE provides a framework for authors to arrive at a quality rating of the evidence for each outcome domain of interest that incorporates information regarding study design, risk of bias, imprecision, inconsistency, indirectness, and magnitude of effect (Guyatt et al., 2011).

In using GRADE, a rating of high quality indicates that authors are “very confident that the true effect lies close to that of the estimate of the effect.” A rating of moderate quality designates that “the true effect is likely to be close to the estimate of the effect, but there is a possibility that it is substantially different.” A low-quality rating specifies that “the true effect may be substantially different from the estimate of the effect”, while a very low-quality rating indicates that “the true effect is likely to be substantially different from the estimate of effect” (Balshem et al., 2011).

GRADE guidelines are such that RCTs begin with a high quality rating while NRS begin with a low-quality rating. Each rating is then modified upward or downward, depending on factors such as risk of bias (including publication bias), imprecision (i.e., large confidence intervals), inconsistency (i.e., heterogeneity in study results), indirectness (i.e., patient population studied differs from the patient population of interest), and magnitude of effect.

Data Synthesis
A narrative synthesis framework was selected for this review rather than a meta-analysis. There was considerable heterogeneity in study design and outcome assessment as well as limited report of inferential statistics; thus, results from individual treatment studies could not be meaningfully pooled. Likewise, a meta-analysis would have been underpowered and difficult to interpret given the limited number of studies with common assessment of outcomes.

Results
Results of the Search
The original search of CENTRAL, MEDLINE, and PsycINFO identified 4,944 articles. After removing duplicates, 1,198 remained. Abstracts of the 1,198 articles were reviewed and 1,141 were excluded. Fifty-seven full-text articles were assessed for eligibility, and 15 studies met the preassigned inclusion criteria. Two of these studies (Douglas & Harris, 2001; Haywood & McCann, 2009) were judged to have exceedingly high risks of bias related to study design, the use of nonstandardized outcome measures, and attrition. As well, many of the authors’ claims regarding the effectiveness of the intervention were not supported by the data presented in the studies. Given these limitations, these studies were deemed to have too many threats to their validity to draw meaningful conclusions and were excluded from further discussion. Thus, 13 studies were retained for the systematic narrative review (Figure 1).

Study Design Characteristics
Two of the 13 studies that met inclusion criteria were RCTs. The remaining 11 NRS were before-and-after designs with no comparison group.

Study Nationality Characteristics
One of the studies was conducted in Austria, one of the studies was conducted in the Netherlands, one of the studies was conducted in Canada, and the remaining 10 studies were conducted in the United States.

Participants
For the RCTs, mean age of participants in the Benoit study (2000) was 17.2 months, while mean age of participants in the Sharp study (2013) was 67.8 months. Mean age of
participants in the NRS was 40.6 months ($SD = 15.1$), and ages ranged from 15.7 to 69.0 months. All of the included studies reported feeding problem or feeding disorder as a primary diagnosis. Three studies specified autism, nine studies specified dependence on supplemental tube feeding, and one study specified spastic diplegic cerebral palsy. Among the 13 studies, 5 studies cited additional medical issues or complex medical history. Three studies targeted child variables alone, and 10 studies targeted both parent and child variables. Mean sample size for the RCTs was 47 ($SD = 24.0$) and ranged from 30 to 64 participants. Mean sample size for the NRS was 54.5 ($SD = 62.5$) and ranged from 9 to 221 participants.

**Types of Intervention**

Many of the included studies reported the use of multicomponent intervention. As such, the following categories of intervention are not mutually exclusive:

1. Behavioral intervention. Behavioral intervention components included stimulus control procedures, extinction, systematic desensitization, shaping, contingency management, and differential attention (11 studies; Benoit et al., 2000; Byars et al., 2003; Clawson, Kuchinski, & Bach, 2007; Cornwell, Kelly, & Austin, 2010; Greer, Gulotta, Masler, & Laud, 2008; Kindermann et al., 2008;
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2. Nutritional intervention. Nutrition intervention components included nutrition education, manipulation of tube feedings, other appetite manipulation, and structured mealtime scheduling (seven studies; Benoit et al., 2000; Byars et al., 2003; Cornwell et al., 2010; Davis, Bruce, Mangiaracina, Schulz, & Hyman, 2009; Kindermann et al., 2008; Silverman et al., 2013; Trabi, Dunitz-Scheer, Kratky, Beckenbach, & Scheer, 2010).

3. Oral-motor intervention. Oral-motor intervention components included oral-motor exercises before meals and oral-motor exercises outside of meals (five studies; Clawson et al., 2007; Cornwell et al., 2010; Greer et al., 2008; Kindermann et al., 2008; Laud et al., 2009).

4. Other psychological intervention. Other psychological interventions included play therapy, family therapy, and psychoeducation (three studies; Kindermann et al., 2008; Laud et al., 2009; Trabi et al., 2010).

5. Caregiver teaching. Studies specified caregiver teaching as an explicit component of intervention (10 studies; Benoit et al., 2000; Byars et al., 2003; Clawson et al., 2007; Cornwell et al., 2010; Greer et al., 2008; Kindermann et al., 2008; Sharp et al., 2011, 2013; Silverman et al., 2013; Trabi et al., 2010).

Effects of Interventions

Detailed outcomes for each primary study are reported in Tables 3 through 17 in the Supplementary Material. Outcomes for each study are summarized in Table I.

Outpatient Interventions

A study was considered to evaluate an outpatient intervention if the frequency of sessions was no greater than once weekly. Three studies were conducted in outpatient treatment centers, with two conducted as RCTs and one as a NRS. Mean duration of intervention was 9.7 weeks ($SD = 3.1$) and ranged from 7 to 14 weeks.

One study (Davis et al., 2009) used appetite manipulation (via tube weaning, defined for the purposes of this article as decreasing supplemental tube feeding in advance of other intervention, and medication management) paired with low-dose medications to reduce pain. A second study (Sharp et al., 2013) evaluated a weekly group behavioral intervention program designed specifically for children with autism spectrum disorders. Benoit and colleagues (2000) combined the two intervention components, implementing a treatment program that paired tube weaning with behavioral therapy.

Tube feedings were successfully eliminated in 100% of the patients in the Davis study and in 13% of the patients in the Benoit study on completion of the intervention programs (tube feedings were eliminated in 47% of patients in the Benoit study at follow-up). Nutrition outcomes were not reported in the Sharp study. With regard to mealtime behavior, no changes were observed in the two studies that evaluated outcomes in this domain (Benoit et al., 2000; Sharp et al., 2013). Finally, decreases in parental stress were observed in the Sharp study.

The findings of these studies demonstrate that in an outpatient setting, interventions composed of appetite manipulation and behavioral therapy are efficacious in transitioning children from tube feeding dependence to oral feeding; however, the relative contributions of each treatment component remain unclear. Furthermore, these outcomes suggest that for a subgroup of patients, outpatient intervention may decrease the need for more intensive intervention.

Day Hospital Interventions

For the purposes of this article, an intervention was considered to take place in a day hospital environment if treatment occurred daily for at least 5 days and patients were not admitted as hospital inpatients. Three of the included studies (Clawson et al., 2007; Sharp et al., 2011; Williams et al., 2007) were conducted in a day hospital environment, and mean duration of intervention was 30.7 days ($SD = 7.6$) and ranged from 24 days to 39 days. Each of these studies was conducted as a before-and-after evaluation of outcomes with no comparison group. All of the included studies evaluated interventions that were primarily behavioral in nature, comprised largely of contingency management components.

Each study reported improvements in disruptive mealtime behavior as evaluated via parent report or direct observation. As well, tube feeding dependence was eliminated for 67% of the participants in the Williams study on completion of the intervention program (tube feedings eliminated for 74% of patients at 2-year follow-up), and participants in the Clawson study demonstrated decreased dependence on supplemental tube feeding on completion of the treatment program. Together these studies support promising treatment effects in the outcome domains of both child mealtime behavior and nutrition when behavioral intervention is conducted in a day treatment environment.
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<tr>
<th>Study (study design)</th>
<th>Study duration</th>
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<th>Intervention</th>
<th>Outcome</th>
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<td><strong>Outpatient</strong></td>
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<tr>
<td>Benoit et al., 2000 (RCT)</td>
<td>7 intervention sessions with follow-up</td>
<td>64</td>
<td>Intervention: nutrition counseling (tube weaning) plus behavioral intervention</td>
<td>No change in mealtime behavior</td>
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<td>Comparison: nutrition counseling (tube weaning)</td>
<td>Less time to tube weaning for intervention group</td>
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<td>At discharge, 13% of patients weaned from tube feeding</td>
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<td>At follow-up, 47% of patients weaned from tube feeding</td>
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<td>Greater proportion of calorie needs met through oral feeding for intervention group</td>
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<td>At discharge, 100% of patients weaned from tube feeding</td>
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<td></td>
<td>At follow-up, 89% of patients weaned from tube feeding</td>
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<td>Davis et al., 2009 (NRS)</td>
<td>14 weeks</td>
<td>9</td>
<td>Appetite manipulation through medication management and tube weaning plus pain rehabilitation</td>
<td>No changes in mealtime behavior</td>
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<td>No changes in dietary variety</td>
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<td>Significant reduction in parental stress for intervention group</td>
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<td>High rates of satisfaction reported by caregivers in intervention group</td>
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<tr>
<td>Sharp et al., 2013 (RCT)</td>
<td>8 intervention sessions</td>
<td>19</td>
<td>Intervention: behavioral intervention</td>
<td>No changes in mealtime behavior</td>
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<td>Comparison: no treatment (waiting list)</td>
<td>No changes in dietary variety</td>
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<td>Significant reduction in parental stress for intervention group</td>
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<td>High rates of satisfaction reported by caregivers in intervention group</td>
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<td><strong>Day treatment</strong></td>
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<td>Clawson et al., 2007 (NRS)</td>
<td>29 intervention days plus follow-up</td>
<td>8</td>
<td>Behavioral plus oral motor intervention</td>
<td>Improved mealtime behavior</td>
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<td>Increased volume of food consumed</td>
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<td>Increased caloric consumption</td>
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<td>Decreased reliance on tube feeding</td>
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<td>Sharp et al., 2011 (NRS)</td>
<td>39 intervention days plus follow-up</td>
<td>13</td>
<td>Behavioral intervention</td>
<td>Improved mealtime behavior</td>
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<td>Increased variety of foods consumed</td>
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<td>Maintenance of gains on parent training and on follow-up at 1 year</td>
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<td>Williams et al., 2007 (NRS)</td>
<td>24 intervention days plus follow-up</td>
<td>46</td>
<td>Behavioral intervention</td>
<td>At discharge, 67% of patients weaned from tube feedings</td>
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<td>At 1 year, 63% of patients weaned from tube feedings</td>
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<td>At 2 years, 74% of patients weaned from tube feedings</td>
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<td><strong>Inpatient</strong></td>
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<td>Byars et al., 2003 (NRS)</td>
<td>11 intervention days plus follow-up</td>
<td>9</td>
<td>Tube weaning plus behavioral intervention</td>
<td>Increased oral caloric intake</td>
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<td>At discharge, 44% of patients weaned from tube feeding</td>
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<td>At follow-up, 67% of patients weaned from tube feeding</td>
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<td>Cornwell et al., 2010 (NRS)</td>
<td>46 intervention days</td>
<td>40</td>
<td>Behavioral intervention plus oral motor therapy</td>
<td>Increased oral caloric intake</td>
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<td>At discharge, 43% of patients weaned from tube feeding</td>
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<td>Greer et al., 2008 (NRS)</td>
<td>Not reported</td>
<td>121</td>
<td>Behavioral intervention plus oral motor therapy</td>
<td>Improved mealtime behavior</td>
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<td>Increased volume of food consumed</td>
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<td>Decreased parental stress</td>
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<tr>
<td>Kindermann et al., 2008 (NRS)</td>
<td>17 intervention days plus follow-up</td>
<td>10</td>
<td>Tube weaning plus psychological intervention plus speech and occupational therapies</td>
<td>At discharge, 100% of patients weaned from tube feeding</td>
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<td>At 6-month follow-up, 80% of patients weaned from tube feeding</td>
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</table>
Inpatient Interventions

Seven of the included studies were conducted in an inpatient environment. Each of these studies was conducted as a before-and-after evaluation of outcomes with no comparison group. Four of the seven studies (Byars et al., 2003; Kindermann et al., 2008; Silverman et al., 2013; Trabi et al., 2010) used appetite manipulation via tube weaning as a primary component of intervention in conjunction with other behavioral or psychological interventions, while the remaining three studies (Cornwell et al., 2010; Greer et al., 2008; Laud et al., 2009) used predominantly behavioral interventions. Mean length of admission was 25.8 days ($SD = 16.7$) and ranged from 11 to 47 days.

Tube feedings were successfully eliminated on completion of the treatment program in 43–100% of patients in the five studies that evaluated nutritional outcomes (Byars et al., 2003; Cornwell et al., 2010; Kindermann et al., 2008; Silverman et al., 2013; Trabi et al., 2010). Improved mealtime behavior was observed in the three studies that specifically evaluated outcomes in this domain (Greer et al., 2008; Laud et al., 2009; Silverman et al., 2013). Greater percentages of patients transitioned from tube feeding to oral feeding in the two studies that primarily used tube weaning with psychological intervention described as a secondary component (Kinderman et al., 2008; Trabi et al., 2010) than in the two studies using tube weaning paired with behavioral intervention (Byars et al., 2003; Silverman et al., 2013). However, mean length of stay was longer with a wider range of treatment duration for the primarily tube weaning interventions, making it difficult to draw conclusions regarding the relative contributions of tube weaning and behavioral intervention as well as conclusions regarding the influence of treatment duration.

Risk of Bias in Included Studies

Risk of bias was assessed for the 15 originally identified studies following the Cochrane Collaboration methodology for systematic review using the two procedures described in the methods section above. In evaluating the design features of each study, the risk of bias was judged to be low for the two RCTs, whereas the risk of bias was rated high for the NRS (Tables 18 and 19 in the Supplementary Material). Specifically, none of the included NRS used a comparison group, putting the findings of the studies at risk for threats to validity, particularly subject to maturation of participants and regression to the mean. As well, 7 of the 13 NRS did not prospectively generate testable hypotheses.

Figures 2 and 3 summarize the results of the bias assessment conducted using the modified version of the Cochrane Risk of Bias Tool. Detailed results of the bias assessment for each individual study are included in Tables 3 through 17 in the Supplementary Material.

In sum, the two RCTs demonstrated an overall low risk of bias. Two of the NRS were removed from further analyses owing to high risks of bias (Douglas & Harris, 2001; Haywood & McCann, 2009). The following is a detailed accounting of specific bias risks identified in the original 13 NRS.

Selection Bias

The identified NRS share the common risk of selection bias, which poses a threat to validity for NRS, particularly...
those that do not use a comparison group (Byars et al., 2003; Clawson et al., 2007; Cornwall et al., 2010; Davis et al., 2009; Douglas & Harris, 2001; Greer et al., 2008; Haywood & McCann, 2009; Kindermann et al., 2008; Laud et al., 2009; Sharp et al., 2011; Silverman et al., 2013; Trabi et al., 2010, Williams et al., 2007). It is possible that specific characteristics of the selected patients contributed to observed improvements, rather than characteristics of the intervention itself. For example, in each of the included studies, criteria for referral to intervention are not reported in detail, and the conditions for referral likely differ from study to study. The decision to treat may be influenced by specific factors (e.g., clinician preferences, patient characteristics, clinical history) that are related to treatment outcome, introducing a source of systematic bias that ultimately leads to an overestimation of treatment efficacy (Deeks et al., 2003). Not only does selection bias present as a potential confound limiting conclusions that can be drawn regarding causality, it precludes synthesis of findings across studies.

Detection Bias
Detection bias was identified in three studies (23%; Douglas & Harris, 2001; Trabi et al., 2010; Williams et al., 2007) and was unclear in two additional studies (15%; Haywood & McCann, 2009; Kindermann et al., 2008). In each of the studies with a high risk of detection bias, outcome assessment was not blinded, and outcome assessment was felt to be influenced by the evaluator’s knowledge of intervention status. For example, in these studies, treatment duration was not prespecified; rather, treatment continued until clinical improvements were observed. As such, outcomes were directly influenced by the evaluators’ awareness of the progression of intervention, resulting in detection bias.

Another potential source of detection bias stemmed from authors failing to report whether the study was conducted as a retrospective chart review or a prospectively designed intervention study, in which hypotheses are
generated and outcome measures selected before implementing the intervention.

Attrition Bias
Attrition bias was identified in five studies (38%; Douglas & Harris, 2001; Haywood & McCann, 2009; Laud et al., 2009; Sharp et al., 2011; Silverman et al., 2013) and was unclear in another study (7%; Byars et al., 2003). Attrition rates averaged 40% and ranged from 10 to 75%. In these cases, not only was attrition high but differences between participants with complete data and those who did not complete final outcome measures could account for study findings. For example, in one study, a high degree of treatment satisfaction was reported on completion of intervention; however, only 30% of the original participants completed this outcome measure (Laud et al., 2009). It is possible that participants who were not satisfied with intervention did not complete the intervention or may have refused to complete postassessment outcome measures.

Reporting Bias
Reporting bias was identified in seven studies (54%; Clawson et al., 2007; Douglas & Harris, 2001; Haywood & McCann, 2009; Kindermann et al., 2008; Sharp et al., 2011; Trabi et al., 2010; Williams et al., 2007). A common example of reporting bias was the use of nonstandardized outcome measures. As well, incomplete reporting of statistical analyses was identified in several studies. Finally, many authors drew conclusions that were not substantiated by data collected or statistical analyses reported.

Other Bias
Other risks of bias were identified in each of the 13 NRS and were largely related to study design (e.g., lack of a comparison group putting studies at risk for threats to validity). These were also identified by the first bias assessment method and are reported above and in the Supplementary Tables 18 and 19.

Quality of the Evidence
Findings of primary studies were evaluated using GRADE methodology (Table II). An overall rating was assigned for each of the following outcomes of interest: observed mealtime behavior, parent report of mealtime behavior, percent dependence on tube feeding, complete wean from tube feeding, grams of food consumed per meal, calories consumed per meal, and parent stress. In using the GRADE approach, observational studies start as low-quality evidence. Thus, the initial quality rating for each outcome was determined to be low, as the majority of the studies were NRS. Low quality is defined by the GRADE Working Group to mean that the true effect may be substantially different from the estimate of the effect (Guyatt et al., 2011).

As per GRADE recommendations, ratings were further influenced by risk of bias, inconsistency, indirectness, imprecision, and publication bias (Guyatt et al., 2011). This subsequent examination of reasons to rate the quality of evidence up or down resulted in rating down the quality of evidence further to very low. Specifically, almost all ratings were influenced by limitations related to study design and small sample size. Additionally, heterogeneity in study findings was identified for four of the outcome domains.

Discussion
The purpose of this systematic review was to summarize and evaluate the best available evidence in support of psychological interventions for pediatric feeding problems. In addition to updating previous reviews to include the most recently available studies and ongoing research, this review outlines the methodological limitations of current primary and review studies, considers the implications that these results may have on clinical practice, and establishes a clear progression for future research.

This review represents a departure from prior reviews, as we included only studies that used between-groups or within-groups comparisons. This is an important advancement, as much of the existing literature is composed of studies using single-subject design. While single-subject design methodologies are well suited to studying specific therapeutic techniques and the effects of modifying or withdrawing treatment, findings cannot be generalized to the larger patient population.

After thorough review of the literature, we identified only 15 studies that met our inclusion criteria of either using RCT methodology or NRS that examined outcomes within a group of participants over time. The limited number of studies identified raises concerns about the slow progression of the literature (since Kerwin completed the first comprehensive review in 1999 the literature has grown on average by one RCT or NRS per year). Of further concern is that only two RCTs were identified. As RCTs are considered the gold standard for evaluating treatment efficacy, only limited conclusions can be drawn from the existing literature regarding the efficacy of behavioral treatments for pediatric feeding problems. However, the identified studies do represent the best available evidence and are summarized here.
Of the included studies, those that used RCT methodology were judged to have the lowest risk of bias. Each of the RCTs was conducted in an outpatient setting and examined outcomes related to child behavior and nutrition. While the target populations and treatment components differed slightly, both studies evaluated the efficacy of behavioral techniques, with Benoit evaluating the additional component of appetite manipulation. Although neither study found directly observable changes in children’s mealtime behavior, Benoit and colleagues found that the combination of appetite manipulation via scheduled tube reductions and behavioral therapy was efficacious in weaning children from supplemental tube feeding (47% of participants able to wean by 4 months posttreatment). When considering the findings of the two RCTs, the multidisciplinary care model used by Benoit may, in part, account for the difference in results lending support for the use of multidisciplinary care models for the treatment of feeding problems.

Further support for a multicomponent treatment approach was found in the 13 identified NRS. Each of these studies was conducted as a before-and-after study with no comparison group. Two studies reported only descriptive statistics, range in percent decrease was 28–65%; variable length of intervention; variable improvement on follow-up.

Table II. Summary of Findings

<table>
<thead>
<tr>
<th>Outcomes</th>
<th>Number of participants (studies)</th>
<th>Quality of the evidence (GRADE)</th>
<th>Comments</th>
</tr>
</thead>
<tbody>
<tr>
<td>Observed mealtime behavior</td>
<td>188 (four studies)</td>
<td>Very low a,b</td>
<td>One study reported only descriptive statistics</td>
</tr>
<tr>
<td>Parent report of mealtime behavior</td>
<td>254 (four studies)</td>
<td>Very low a,b,c</td>
<td>Two studies reported only descriptive statistics, range in percent decrease was 28–65%</td>
</tr>
<tr>
<td>Percent dependence on tube feeding</td>
<td>94 (three studies)</td>
<td>Very low a,b,c</td>
<td>Range in percentage of patients weaned was 13–100%; variable length of intervention, variable improvement on follow-up</td>
</tr>
</tbody>
</table>
| Tube dependent (Y/N)            | 444 (eight studies)             | Very low a,c                    | **Note. RCT = randomized controlled trial; GRADE = Grading of Recommendations Assessment, Development, and Evaluation; NRS = nonrandomized studies.
GRADE Working Group grades of evidence: High quality ◆◆◆◆◆: We are very confident that the true effect lies close to that of the estimate of the effect. Moderate quality ◆◆◆◆◆◆: The true effect is likely to be close to the estimate of the effect, but there is a possibility that it is substantially different. Low quality ◆◆◆◆◆◆: The true effect may be substantially different from the estimate of the effect. Very low quality ◆◆◆◆◆◆: The true effect is likely to be substantially different from the estimate of effect. Assessments were made using the GRADE guidelines for rating the quality of the evidence as follows (Guyatt et al., 2011):
aLimitations due to study design and implementation and associated risks of bias.
bImprecision of results due to small samples sizes.
cInconsistency (heterogeneity in study results).**
implemented in six of the NRS (54.5% of the included NRS) and was used in one outpatient setting and in five of the inpatient hospital settings. Improved nutritional outcomes were reported in each of these studies.

Neither of these intervention components was implemented in isolation; rather, each of the studies reported some degree of multidisciplinary care. Other interventions used but not explicitly evaluated or identified in hypotheses included caregiver training (72.7% of the studies), oral-motor therapy (45.5% of the studies), and other forms of supportive therapy (27.3% of the studies). The frequency with which these additional intervention components are identified in these 13 studies further supports the use of multicomponent care for the treatment of pediatric feeding problems. Given the complexities associated with the delivery of interdisciplinary care it is not surprising to see that the greatest number of studies that reported implementation of both behavioral and nutritional intervention components occurred in inpatient settings.

While the results of these NRS are promising, caution must be exercised in drawing causal conclusions regarding the efficacy of these interventions. Because none of these studies used a comparison group, conclusions drawn by the primary authors are limited by threats to validity, specifically maturation effects and the possibility that treatment effects could be explained by regression to the mean. This is more pronounced when treatment duration is permitted to vary according to the interventionist’s assessment of clinical progress. However, these studies do represent the best available evidence and provide support for the continued use of behavioral intervention for the treatment of pediatric feeding problems. The collective work also serves as a foundation for the design of future studies.

Authors’ Conclusions

Implications for Practice

In the absence of RCTs, it is not possible to specify which specific environments, interventions, or intervention components are most efficacious in the treatment of pediatric feeding problems. However, the best available evidence comprised nonrandomized before-and-after studies shows promising outcomes when behavioral interventions that include nutritional manipulation are implemented. In terms of intervention environment, although both RCTs were conducted in an outpatient setting, the efficacy of intervention was only supported in one. Rather, inpatient and day treatment programs have the most available support for positive treatment outcomes, supporting ongoing implementation of behavioral intervention in these environments.

Implications for Research

RCT methodology is considered to be the gold standard to determine the efficacy of an intervention. As such, the RCTs identified in this review are highly valuable in setting an agenda for future research evaluating interventions for feeding problems. These studies demonstrate that the rigor of RCT methodology is possible within this population of patients.

Findings from the NRS identified in this review establish a clear progression for future, more stringent evaluation of interventions. A reasonable next step would be the conduct of prospectively designed controlled before-and-after studies. This can easily be done using a waiting list comparison group. Subsequently, outcomes from controlled before-and-after studies can serve as pilot data for the careful design of RCTs. RCTs can be used to compare behavioral interventions to other forms of intervention (e.g., oral-motor therapy, sensory integration therapy), to evaluate specific components of intervention (e.g., appetite manipulation, behavioral intervention), and to evaluate the effectiveness of intervention in specific environments of care (e.g., outpatient behavioral intervention, inpatient behavioral intervention). Multisite collaboration can facilitate the implementation of this more rigorous research methodology.

Additionally, the use of standardized, multimethod outcome assessment is essential in the design of future intervention studies. Most currently available studies evaluated either mealtime behavior change or nutritional status but not both. As well, a single assessment method was typically used to evaluate outcomes in a particular domain (e.g., parent report of mealtime behavior). Future study using multimethod outcome assessment (e.g., parent report and direct observation of mealtime behavior paired with clinician assessment of nutritional status) can better examine the relationships among parent behavior, child behavior, and nutrition in a methodologically sound manner.

Once more systematic study has been established, the efficacy of individual treatment components can be examined and mediators and moderators of treatment outcome can be identified, such that treatment can be altered to maximize efficiency. For example, findings of future study may lead to the ability to tailor treatment to particular patient populations. This may then lead to decreased duration of intervention, lessening the cost of intervention. Many of the authors of the included primary studies discuss the cost-effectiveness of behavioral intervention; however, much of the discussion at this time appears to be speculative rather than systematically studied. Methodologically sound evaluation of cost-effectiveness warrants future attention as well.
Difficulty in Conducting Primary Studies
The conduct of efficacy research with the population of pediatric feeding problems is remarkably difficult for a number of reasons. The heterogeneity in the manifestation of feeding problems (e.g., food selectivity, failure to advance to a developmentally appropriate diet, growth problems, under nutrition) paired with the variability in patient characteristics (e.g., five of the studies describe patients with complex medical history, three specify autism as a primary diagnosis, ages range from 15 to 70 months) presents a unique challenge when selecting study design. The majority of included studies were conducted in an intensive treatment environment, which also suggests the complexity of these problems. Despite this degree of complexity, treatment outcomes are positive and promising.

It is noteworthy that most of the programs evaluated use multidisciplinary interventions. The preliminary evidence suggests positive effects of multidisciplinary treatment; however, no identified studies have conducted a component analysis. At this point, as interdisciplinary intervention has become the standard of care, it may be difficult to remove any particular component, as this would represent a departure from current standards of care. However, future research that is conducted in a careful and systematic manner can effectively evaluate these components without sacrifice to patient care.

Conclusions
In sum, the available evidence suggests positive treatment effects of psychological interventions for pediatric feeding problems. However, significant methodological limitations preclude firm conclusions regarding the efficacy of behavioral intervention for pediatric feeding problems. Future studies that use more rigorous research methods are needed. To enhance our understanding of these types of treatments, studies that reduce the risks of bias observed in this review are urgently needed. Future work should include prospective RCTs of protocolled interventions. Studies should include longitudinal data whenever feasible. Component analyses studies are also needed to best understand the relative contributions of interdisciplinary treatments.

Supplementary Data
Supplementary data can be found at: http://www.jpepsy.oxfordjournals.org/

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References


