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IMPROVING NUTRITION IN TODDLERS AND PRESCHOOL CHILDREN WITH CYSTIC
FIBROSIS: BEHAVIORAL PARENT TRAINING INTERVENTION

A dissertation submitted in partial fulfillment of the requirements for the degree of
Doctor of Philosophy at Virginia Commonwealth University

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Abstract

IMPROVING NUTRITION IN TODDLERS AND PRESCHOOL CHILDREN WITH CYSTIC FIBROSIS: BEHAVIORAL PARENT TRAINING INTERVENTION

By Shannon E. Hourigan, M.S.

A dissertation submitted in partial fulfillment of the requirements for the degree of Doctor of Philosophy at Virginia Commonwealth University

Virginia Commonwealth University, 2012

Major Director: Michael A. Southam-Gerow, Ph.D.
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The purpose of this single-case study was to pilot a developmentally sensitive adaptation of an evidence-based intervention aimed at improving nutrition in children with cystic fibrosis (CF). Children with CF must adhere to a high-calorie diet to prevent malnutrition and maintain health. Toddler and preschool age children present a unique feeding challenge to parents as they begin to exert independence and exhibit a variety of challenging behaviors. Parents trying to adhere to CF nutrition guidelines often use ineffective strategies that inadvertently encourage children *not* to eat. This six-week group parent-training intervention combined nutrition and behavior elements to provide parents with the nutrition and child behavior management strategies necessary to improve children's intake and ensure adequate nutrition.

Parents of four children (one girl) between the ages of 21 and 30 months of age participated in two groups; all children were Caucasian, and all parents were married. Two children were malnourished and had primary goals of increasing intake and weight; two were

adequately nourished and had primary goals of improving diet quality. Primary and secondary treatment outcomes were established individually for each child. Families completed three-day diet diaries and video recorded mealtime interactions across two baseline weeks, six weeks of intervention, and a twelve-week post-intervention follow-up. Children's weights were measured at baseline, post-treatment, and follow-up. Caloric intake was calculated for all meals, and video taped meals were coded using a behavioral coding system. Treatment fidelity was also assessed.

The two malnourished children increased caloric intake throughout the intervention and demonstrated clinically significant weight gains at post-treatment. Further, these gains were maintained at follow-up. One of the two adequately nourished children demonstrated improved diet quality. Findings support the efficacy of this developmentally-sensitive adaptation to increase weight in toddler children with CF, and findings provide partial support for the efficacy of this intervention in improving diet quality in adequately nourished children. A parent group intervention provides training in CF-specific child management skills to multiple families and may provide significant benefits to parents who often struggle with the demands of nutrition requirements and toddler behavior.

Improving Nutrition in Toddlers and Preschool Children with Cystic Fibrosis:

Behavioral Parent Training Intervention

Review of the Literature

Cystic Fibrosis (CF) is a chronic, genetic, life-limiting disorder that affects approximately 30,000 children and adults in the United States (Cystic Fibrosis Foundation [CFF], 2007). The focus on research in past decades has resulted in an increase in the median life expectancy from 25 years in 1985 to 37 years in 2007 (CFF, 2007). Although CF is no longer exclusively a disorder of childhood, 25% of deaths occur in children, adolescents, and young adults under the age of 21 (Zemanick et al., 2010). CF impacts multiple organ systems, and disease management focuses on slowing the progression of the disease and requires individuals to complete complex treatment regimens that involve over an hour of treatment related activities each day (Orenstein, 2004; Ziaian et al., 2006). One critical treatment element involves adherence to a high calorie diet to maintain adequate body weight. This paper first reviews the literature demonstrating the importance of nutrition in CF treatment, then presents the rationale for a study designed to test a psychosocial intervention to improve dietary intake and behaviors at meal time for toddlers and preschool children with CF.

The literature review will achieve four aims. First, it will provide a brief overview of the pathophysiology of CF, second, it will turn to a discussion of disease management, focusing on the complex treatment regimens required of families and the issue of adherence. Third, it will review the impact of nutrition on the progression of the disease and the importance of ensuring

adequate nutrition in toddler and preschool children; finally, it will review the behavioral nutrition intervention literature to identify the gaps that will be addressed by the proposed study.

Pathophysiology

Cystic Fibrosis is a serious, life-limiting, genetic disorder that affects respiratory, digestive, reproductive and endocrine systems. It is the most common terminal genetic disease among Caucasian populations, affecting approximately 1 in 3,400 live births (CFF, 2007; FitzSimmons, 1993; Quittner, Modi & Roux, 2004). However, it is far less common in non-Caucasian populations, affecting approximately 1 in 17,000 African Americans and 1 in 90,000 Asian Americans in Hawaii (Orenstein, 2004). CF is a recessive autosomal disorder, thus both parents must pass a copy of a CF gene to their child for CF to be present, and there are many different alleles that can produce CF (Quittner et al., 2004; Castellani et al., 2008). CF is typically diagnosed in infancy (CFF, 2007; Stark, Mackner, Patton & Acton, 2003), and forty-eight states include CF in the newborn genetic screening panel (Zemanick et al., 2010), improving opportunities for early intervention. Intensive investment in research of CF treatments has resulted in marked improvements in life expectancy. In 1974, median life expectancy was 8 years; since 1985, median survival age has increased from approximately 26 years to 37.4 years (CFF, 2007). Similarly, the proportion of adults in the CF population has steadily increased, and people age 18 years and older currently account for approximately 45% of all patients (CFF, 2007).

Pulmonary effects. The basic defect in CF involves passage of chloride ions through cellular walls. One outcome of this defect is increased salt excretion from the sweat glands. Indeed, identification of children with CF in the past often followed parents noticing a salty taste when kissing their infants (Orenstein, 2004). This defect also results in the production of sticky

mucus within the lungs and other organs with secretory functions. Unlike the thinner mucus that forms a moving layer in lungs of healthy individuals and aids in the expulsion of foreign substances, the thick, viscous mucus of individuals with CF accumulates in the lungs and restricts airflow to the tiny sacs (alveoli) where gases are exchanged with the bloodstream. In addition to clogging airways, this mucus provides a hospitable host environment for bacteria and viruses that are thought to cause inflammation that results in tissue damage (Flume et al., 2009; Orenstein, 2004). Further, byproducts of this inflammation and infection contribute to the viscosity of CF mucus as the DNA from the white blood cells released by the body to fight airway infection collect in the mucus, break down, and add to the thickness (Flume et al., 2009; Orenstein, 2004). In this way, the body's defensive response to infection contributes to the creation of an environment conducive to that infection.

Treatment for CF aims to clear the airways of the continually forming and increasingly trenchant mucus by performing various airway clearance techniques to shake the mucus out of the small airways and into the upper airways (bronchi) where the mucus can be coughed out of the lungs. One technique called percussion, commonly as chest physiotherapy (CPT), includes clapping firmly on the chest to shake the mucus out of the lung (Orenstein, 2004; Samuels, 2000). In addition to CPT by hand, devices such as mechanical percussors that provide mechanical pounding or vibrating vests are also used to shake mucus out of the lungs. The goal of all of these treatments is to postpone and minimize the impact of the chronic cycle of inflammation, infection, and lung damage that occurs when lung tissue scarred by inflammation becomes ineffective at clearing mucus (Flume et al., 2007, 2009; Orenstein, 2004; Quittner et al., 2004). Despite the best efforts of patients, families, and treatment teams, respiratory failure accounts for approximately 85% of deaths (Flume et al., 2009; Quittner et al., 2004).

Given the centrality of lung health to disease severity in CF, measures of pulmonary functioning are critical indicators in the evaluation of health. There are three primary indicators of pulmonary function. Forced vital capacity (FVC) is the volume of air that is blown out in a single breath. Forced expiratory volume at 1 second (FEV₁) is the amount of air that is blown out in the first second of the FVC. FVC is a measure of overall lung capacity and FEV₁ is an indicator of airway obstruction used to measure decline in pulmonary functioning (Corey, Levison & Crozier, 1976 as cited in Stark et al., 1993; Orenstein, 2004). Percent of FEV₁ predicted by height, sex, and race is often used as a measure of lung functioning. For reference, a healthy individual without any lung disease would be expected to demonstrate 100% FEV₁ predicted, and when FEV₁ falls below 30%, that patient is thought to have a 50% chance of survival in two years. However, it is important to note that FEV₁ is effort-dependent, meaning that an accurate measurement requires the patient try the best that they possibly can, and it is a measure that may fluctuate as a function of current infection and inflammation (Orenstein, 2004), thus treatment providers typically look at trends in FEV₁ predicted.

Pancreatic effects. In addition to the pulmonary impact of the CF mutation, approximately 90% of individuals with CF require pancreatic supplements to replace digestive enzymes prevented from secretion by mucus related blockages (Borowitz, Baker & Stallings, 2002; CFF, 2007). Untreated or inadequately treated, pancreatic insufficiency can result in malabsorption of nutrients, malnutrition, poor growth, and digestive difficulties such as abdominal discomfort and loose, greasy stools (Borowitz et al., 2002). Thus, most individuals with CF must take replacement enzymes with every snack and meal to adequately digest and absorb nutrients. The impact of CF is far more extensive than described in this cursory review.

Interested readers should consult Orenstein (2004) for well-written coverage of this complex disease.

CF patients are prescribed a great many more treatments than replacement enzymes and airway clearance. For these treatments to be effective, patients must adhere to the treatment as prescribed. The next section will review some of the commonly prescribed treatments for CF to illustrate the complex and time consuming requirements of managing this disease.

Treatment Regimen

The complexity of CF disease and the importance of airway clearance on survival contribute to a complicated and intense treatment regimen that contributes greatly to long-term health and survival (Modi & Quittner, 2006; Quittner et al., 2004). However, typical treatments for children with CF are complex and extremely time consuming, even when children have a very mild disease. This review will discuss several common elements of treatment with attention paid to what comprises each treatment, the evidence in support of the treatment, and its burden on families. The treatments described below are all prescribed in addition to the multitude of medications that must be taken orally on a regular basis.

Airway clearance. The sticky mucus found in the lungs of individuals with CF obstructs the small airways, provides a hospitable environment for bacterial infection, and contributes to a chronic cycle of inflammation and injury to lung tissue (Flume et al., 2009). This chronic cycle of lung infection and inflammation results in irreparable damage to lung tissue that contributes to death in 85% of individuals with CF (Flume et al., 2009). Given the importance of airway clearance in the maintenance of health, it is not surprising that a number of techniques exist purporting to clear the airways of mucus.

There are several techniques that are frequently recommended to assist in the clearance of airways. This review will cover those recommended for use in young children; interested readers are referred to Flume and colleagues (2009) for further details and information about other airway clearance methods. Three techniques are commonly used for airway clearance in young children: percussion and postural drainage, high-frequency chest compression, and exercise. Percussion, also known as CPT, requires the parent or caregiver to clap firmly on the child's chest or back to shake the mucus loose. This treatment can be time consuming for both patients and caregivers, requiring 15-30 minutes for each session (Orenstein, 2004; Samuels, 2000). Research examining sputum production and lung function demonstrated that CPT is efficacious in clearing sputum from airways and maintaining lung function (Flume et al., 2009). High-frequency chest compression (HFCC) is another commonly used airway clearance strategy. HFCC purports to vibrate the airways and loosen mucus with pulses of pressure in an inflatable compressive vest that the child wears for 15-30 minutes. The HFCC vest frees parents or caregivers from providing manual CPT, but the equipment is bulky and can be cost prohibitive for some families (costing upwards of \$16,500; Hill-Rom, N.D.; Orenstein, 2004). Neither the HFCC nor exercise has been compared to no-treatment on clinical outcomes, but both have been compared to CPT. When sputum production and lung function are used as the clinical outcomes, the data comparing HFCC and CPT are equivocal (Flume et al., 2009). Physical exercise is also identified as an airway clearance technique. Evidence suggests that CPT alone provides a greater benefit than exercise alone in improving lung function, and physicians and researchers suggest exercise should be used as an adjunctive therapy (Flume et al., 2009). Overall, patient and family preference likely dictates which treatment is most beneficial for an individual family as

preference is likely related to adherence, and neither CPT nor HFCC has consistently demonstrated clinical superiority.

Aerosols. Patients are prescribed a wide variety of inhaled medications. Broadly, these medications serve three functions. The first is to physically alter the mucus to facilitate airway clearance. This is done by either improving hydration of the mucus via inhaled hypertonic saline (solution with a high saline concentration) or by breaking up the mucus via an inhaled enzyme that catalyzes DNA from white blood cells that accumulates in and thickens the mucus. The second function is to combat the bacteria that infect the airways and cause inflammation via the use of inhaled antibiotics. The third function is to dilate the airways in patients who have asthma in addition to CF. These treatments involve the use of a pressurized metered dose inhaler, a dry powder inhaler, or an air compressor machine that turns liquid into a mist that can be easily inhaled into the lungs. Inhaled medication can be delivered in a much higher concentration than oral or intravenous medication and can be delivered directly to the site of inflammation, mucus, or infection (Flume et al., 2007; Orenstein, 2004). When prescribed under careful medical supervision, all three classes of inhaled medications described above have demonstrated clinical effectiveness and have been recommended by the committee established by the CFF to examine the clinical evidence for pulmonary therapies (Flume et al., 2007). Total duration of treatment depends on the number of medications prescribed and the frequency of treatments, but inhaled hypertonic saline, the mucolytic enzyme, a bronchodilator, and an antibiotic are often prescribed in addition to CPT. All can extend the total time for treatments to close to an hour.

Diet and nutrition. The importance of diet and nutrition in the treatment of CF will be covered in more depth later but will be briefly reviewed in this section. Children with CF require more energy than do children without CF to maintain the same weight. Children with CF have a

higher metabolic rate than children without CF due to increased energy required by diseased and damaged lungs coupled with difficulty absorbing nutrients from food due to decreased production of pancreatic enzymes. Thus, children with CF are often faced with an energy imbalance that makes gaining and maintaining normal weight difficult.

Prior to genetic screening, failure to thrive and scrawny appearance were indicators of CF disease (Orenstein, 2004). Evidence demonstrates that nutrition status is very closely related to lung function. Higher BMI percentile (the standard measure of body composition in children) is associated with better lung function (Borowitz et al., 2002; CFF, 2007; Ramsey, Farrell, Pencharz & the Consensus Committee, 1992), and early nutrition status during the toddler and preschool years is predictive of lung function during the school age years (Konstan et al., 2003; Lai, Schoff, Farrell & Wisconsin CF Neonatal Screening Group, 2009). Given the importance of weight and nutrition status in the maintenance of lung health, it is recommended that children with CF consume between 120% and 200% of the recommended dietary allowance (RDA; National Research Council, 1989) of their same age peers with 40% of calories coming from fat (Stallings et al., 2008). Thus, children with CF are required to eat three high-calorie meals and several snacks throughout the day to maintain optimal weight. These demands can place significant strain on families (Powers, Mitchell et al., 2005), and many families have difficulty with adherence to this high-calorie dietary regimen (Powers et al., 2002; Stark et al., 1997; Tomezsko, Stallings & Scanlin, 1992). In more severe stages of disease or malnutrition, the placement of a feeding tube in the gut (the enteral route) may be required (Lester, Rothberg, Dawson, Lopez & Corpuz, 1986) to ensure adequate nutritional intake.

Pancreatic enzymes. In addition to caloric intake at 120%-200% RDA, most CF patients must also take pancreatic enzymes to assist in digestion (Borowitz et al., 2002). The combination

of adequate caloric intake and pancreatic enzymes are key aspects of care in meeting the nutritional needs of children with CF. The efficacy of pancreatic enzymes is well established in the treatment of pancreatic insufficiency for children with CF (Stallings et al., 2008), and following a systematic review of the literature, the CFF Subcommittee on Growth and Nutrition recommends the administration of pancreatic enzyme replacements before meals or snacks containing fats to ensure adequate absorption of fats and nutrients (Stallings et al., 2008). These recommendations require children to take pills containing dietary enzymes before each meal and snack containing fat; however, research found less than 30% of children demonstrate adherence to enzyme therapy (Shall, Bentley & Stallings, 2006), suggesting the majority of children are not fully absorbing the calories and nutrients they consume.

Compliance with the CF treatment regimen requires a significant daily time and energy commitment, and the burdens placed on children and families with CF are significantly higher than those encountered by children suffering from other chronic conditions. In a study examining the treatment burden and associated health-related quality of life in children with CF, type 1 diabetes, and asthma, Ziaian and colleagues (2006) found that children with CF reported spending an average of approximately 74 minutes per day on treatments compared to 57 and 6 minutes per day for children with diabetes and asthma, respectively. For all diseases, parents reported that children spent less time completing treatment than children reported, but parents of children with CF reported significantly more time in treatment (60 minutes) than parents of children with diabetes and asthma (34 and 6 minutes, respectively). Further, there was a significant negative relationship between FEV₁ and treatment duration, indicating that treatment time increased as lung function worsened (Ziaian et al., 2006). Thus, CF is complicated,

cumbersome, chronic, and requires significantly more time than treatment of other childhood chronic illnesses, presenting significant challenges to optimal treatment adherence.

Adherence to Treatment

Adherence to the complex treatment regimen prescribed for disease management is the subject of many scholarly articles and chapters as patients' adherence to prescribed treatments is central to disease management, and nonadherence to treatments impacts not only health care outcomes, but data from scientific studies examining clinical efficacy (Reikert & Drotar, 2000; Quittner, Espelange, Ievers-Landis & Drotar, 2000; Quittner, Modi, Lemanek, Ievers-Landis, & Rapoff, 2008; Stark, 2000). Adherence is an important issue for clinicians and researchers alike, and the goal of improving adherence to chronic pediatric conditions poses unique challenges, one of which is the changing needs of the developing child. Children's developmental level impacts adherence as children's willingness or resistance to engage in treatment varies over time and the responsibility for treatment of this lifelong disease begins with parents and gradually is transferred as the patient matures. Although the issue of transition of treatment responsibility is important, this review will focus primarily on those factors that may be related to adherence to treatment for toddler and preschool children. Interested readers are referred to Creer (2000) or Modi and colleagues (Modi, Marciel, Slater, Drotar & Quittner, 2008) for a discussion of factors associated with transferring responsibility for treatment throughout the lifespan.

Bauman (2000) identifies two different, though not mutually exclusive, types of nonadherence: volitional and involuntary. Volitional nonadherence occurs when a patient hears and understands treatment recommendations but makes a conscious decision not to comply. Involuntary nonadherence occurs when the patient accepts treatment advice and believes they are adhering *well enough* to be considered sufficient. This review will focus on several factors

related to both types of nonadherence, specifically patient knowledge, complexity, burden, and beliefs.

Knowledge. Parents of children with CF must have an accurate understanding of the clinically relevant details in their treatment regimen to be adherent to treatment recommendations. The amount of information required for parents to oversee the day-to-day administration of children's care is staggering. Parents are required to know: the names and doses of prescribed and non-prescribed medications, the frequency of treatments, the duration of treatments (i.e., days to take a medication or minutes to perform treatment), the order of administration (i.e., aerosol medications prior to CPT), and the special techniques required to perform treatments (e.g., how to clean nebulizers, how to determine the correct enzyme dose, how to perform CPT; Ievers-Landis & Drotar, 2000). Without this knowledge, treatment adherence is impossible.

The relationship between global knowledge about CF and adherence suggests that it is not general disease knowledge that predicts adherence but rather understanding specific details of the prescribed individual treatment regimen (Nolan, Desmond, Herlich & Hardy 1986; Quittner et al., 2000). Studies assessing parental knowledge of medical treatments suggest that for the most part, parents do not accurately recall provider recommendations. As an example, in a study of parents of children with CF, 14% of parents reported that CPT was unnecessary if the child was feeling well (Henley & Hill, 1990). Quite the opposite, CPT is required on a daily basis to continually break up sticky mucus in the airways and minimize opportunity for infection. Further, 20% of mothers and 24% of fathers believed that pancreatic enzymes were not necessary outside of the three main meals, when they're required at any meal in which fats are

ingested (Henley & Hill, 1990). Despite the best efforts of treatment providers, a significant portion of parents did not fully understand some key aspects of CF treatment.

The findings of Henley and Hill (1990) are not isolated. Another study found that 47% of parents of children disagreed with their treatment provider about whether a special diet had been prescribed (Gudas, Koocher & Wypij, 1991), when the recommendation for all children with CF is caloric intake of at least 120% of RDA. Yet another study found 92% of parents were unaware that fat has more calories than carbohydrates or protein (Modi & Quittner, 2006), a critical piece of knowledge when families are tasked with getting their child to consume 120% to 200% of the calories expected of their same age peers. Thus, research indicates that there are significant gaps in parents' knowledge of treatment. However, factors contributing to this gap and to the level of nonadherence are not well understood.

One factor thought to contribute to parents' knowledge of treatments is the quality of patient-provider communication (Ivers-Landis & Drotar, 2000). Research comparing the relationships among patient time spent with treatment providers, amount of time physicians spent discussing treatment related issues, and recall of medial information in adult patients demonstrates that time with providers discussing either treatment recommendations or adherence itself increases patient recall of treatment recommendation details in adults (Ivers-Landis & Drotar, 2000). Although this has not been directly tested, it would not be unreasonable to expect that this relationship would extend to parents of children with chronic medical conditions.

Complexity. Knowledge of treatment recommendations is a necessary first step to good adherence; however, masterful understanding of treatment regimen becomes more difficult as treatment complexity increases. Indeed, a study with an adult population identified an inverse relationship between patient knowledge of medications prescribed and the number of

medications prescribed (Fletcher, Fletcher, Thomas & Hamann, 1979). Adherence to CF treatment regimen, including chest physiotherapy, oral and inhaled medications, and dietary regimens is closely associated with long term survival. Adherent patients must complete multiple rounds of CPT, consume a high calorie diet, balance enzyme levels, and administer several medications on different dosing schedules using different routes of administration. This series of complicated steps increases chances for nonadherence (Quittner et al., 2000, 2008). Research examining the relationship between complexity and adherence has demonstrated that rates of adherence to complex regimens such as dietary monitoring and physical therapy are worse than adherence to medications (Reikert & Drotar, 2000). Similarly, Passero and colleagues found that 93% and 90% of patients reported compliance with antibiotic and vitamin therapy, respectively, but only 40% of patients reported adherence to chest physical therapy regimens and only 20% reported adherence to dietary regimens (Passero, Remor & Salomon, 1981). Poor dietary adherence has been reported in other research, with adherence rates ranging from 12% - 16% (Anthony, Bines, Phelan & Paxton, 1998; Eddy et al., 1998; Mackner, McGrath & Stark, 2001). Rates of adherence to treatment are neither optimal nor consistent.

Further complicating understanding of adherence, research demonstrates rates of reported adherence vary by the method of assessment. Modi and colleagues (2006) compared rates of adherence using several assessment methods, parents' and children's self-report, parent report using an evidence-based cued 24-hour telephone-based recall procedure and pharmacy refill data (where applicable). They found that rates of adherence varied widely as a function of the assessment method. For example, rates of reported airway clearance frequency varied from 51.1% when assessed via cued recall to 74.4% via parent self-report. Similarly, adherence rates to enzyme treatment ranged from 46.4% using cued recall to 90.0% and 89.5% via child and

parent self-report respectively. Adherence to daily vitamins varied from 22.2% via cued recall to 88.4% and 93.8% via parent and child report, respectively. Adherence to inhaled dornase alfa ranged from 56.7% via cued recall to 71.7% via pharmacy refill history to 90.4% and 77.8% via parent and child report, respectively. Finally, adherence to inhaled tobramycin (an important antibiotic) ranged from 36.1% via cued recall to 85.0% and 83.3% via parent and child report (Modi et al., 2006). The tremendous variability in adherence rates demonstrates significant inflation in parent and child self-report of adherence when compared to the more objective measures of the 24-hour cued recall procedure and the pharmacy refill history. This inflation may reflect families' wanting to please providers by appearing to follow treatment recommendations and may be evidence of Bauman's (2000) involuntary nonadherence wherein families believe they are completing treatments well enough to be considered sufficient. While many of frequently missed treatment elements (e.g., vitamins) are not complex on their own, these data speak to the challenge presented to families in consistently adhering to the overall treatment regimen.

Clearly, families do not consistently adhere to the treatment regimen prescribed by health care providers. Dietary monitoring and physical therapy are not only complex treatments, but they also require significant investments of time and energy and may be disruptive to the family. Further, the demands on the family are not consistent in that children may experience periodic exacerbations that increase demands on family followed by periods of infrequent symptoms and relative calm (Creer, 2000). Thus, the already complex CF treatment routine is made more complicated by the waxing and waning presentation of symptoms and associated treatment demands.

Burden. Many of the treatments prescribed for children with CF require significant investments of time and energy from both parents and patients. As an example, an adherent CPT routine without the additional burden of inhaled antibiotics requires the patient to first take inhaled or nebulized medications designed to loosen mucus then use either a therapy vest or receive manual CPT from a parent or family member. The amount of time required for both the inhaled medications and PT range from approximately 45-60 minutes, and CPT is generally prescribed at least twice per day, more frequently as the disease progresses. The nebulizer and therapy vest machines are portable – the newest therapy vests come in cases the size of carry-on luggage, but unlike pressurized metered dose inhalers or blood glucose monitors, this equipment is neither discreet nor easily used in public. Thus, it may be difficult for families to complete therapies in the middle of the day or while not at home. Indeed, research looking at adherence in asthmatic preschool aged children indicates that when treatments were prescribed three or four times per day, those doses administered during the middle of the day were most frequently missed (Gibson, Ferguson, Aitchison & Paton, 1995). These findings provide support for the laments often heard in treatment clinics: that families have difficulty meeting the competing demands of family life and children’s treatment routines.

Costs also present a significant burden to families and risk for nonadherence (Bauman, 2000). Average annual out of pocket health care cost for a single privately insured individual with CF is \$3,300 in addition to the cost of health care premiums, with 39% of all health care expenditures attributable to the cost of prescription medications (Ouyang, Grosse, Amendah & Schechter, 2009). Therapy vest machines can cost up to \$16,425 (Hill-Rom), and families who cannot afford a vest must commit between 20 minutes and two hours of adult time per day to ensure children receive adequate manual CPT. Families also must budget for expenditures to

travel to quarterly clinic visits, remain with children during inpatient hospital stays, and obtain food for special diets (Bauman, 2000). Despite the significant financial burdens associated with adherent CF treatment, consistent relationships between adherence and SES have not been demonstrated (Matsui, 2000), indicating that whereas cost may pose significant burdens to families, it is neither the only burden nor is it an insurmountable one.

Child behavior may also serve as an additional burden and barrier to adherence. Oppositional behavior was frequently endorsed by parents in a survey of barriers to adherence with several components of treatment. Fifty percent of parents identified oppositional behavior as a barrier to nebulizer treatments, 60% as a barrier to airway clearance, 39% as a barrier to dornase alfa, 23% to nutrition, and 25% to inhaled antibiotics. Families also frequently endorsed forgetting and time management as common barriers to treatment adherence (Modi & Quittner, 2006). Taken together, there are a number of factors in family life that can present obstacles to reliable treatment completion.

Beliefs. In addition to structural barriers that can present obstacles to treatment completion, the way families think about treatment, disease, and their role in health promotion may contribute to adherence. The choices families make when prioritizing family activities and scheduling treatments are influenced by underlying fears, values, or beliefs about treatment (Bauman, 2000). One such underlying factor is the extent to which a patient or family believes the treatment is helpful, and it is not uncommon for patients to experiment with medication to test its efficacy. This patient skepticism is particularly problematic when the benefits of the medication or treatment are not immediately evident (Bauman, 2000), as is the case with many CF medications and treatments designed to minimize opportunity for infection, reduce mucus, maintain a healthy weight, and keep the airways clear. Indeed, research suggests that adherence

to prophylactic medication is poorer than adherence to medications for acute conditions (Fortheringham & Sawyer, 1995). Thus, when not reinforced by an immediate sign of treatment efficacy, patients are less likely to complete treatments in the future.

CPT is an excellent example of a treatment that does not always have immediately obvious benefits. As described previously, the goal of physiotherapy is to generate vibration to loosen mucus in the small airways. This newly freed mucus is not immediately coughed out by the patient as it first must work its way up the mucocilliary escalator (i.e., the upwards pathway along which mucus and foreign matter are transported out of the lungs) through the small airways and into the bronchi before a cough reflex can be initiated. Thus, despite documented effectiveness of CPT, the lack of immediate signs of benefit may result in belief that CPT is not effective.

Adherence to dietary recommendations may be impacted if families feel conflicted about the messages they receive from the CF care team and information to the general public about the healthfulness of certain foods. Children with CF should consume 120% - 200% RDA, but the message to feed children high calorie, high fat meals conflicts with recommendations given to the general public about reducing intake. Parents may experience dissonance or difficulty giving their children food they believe to be “unhealthy” (e.g., extra butter, fatty “junk foods,” or heavy cream).

Finally, research suggests that cognitive styles and attributions about control are related to adherence. Perception of low internal control about health (i.e., low sense of belief that health can be controlled by one’s own actions) and external attributions about negative events (i.e., the belief that negative events are random and uncontrollable) are associated with decreased adherence in adolescents with Type 1 diabetes (Murphy, Thompson & Morris, 1997). In addition

to patient beliefs, parents' attributions about their children's chronic health conditions are related to health behaviors and adherence. In a study examining metabolic control in children with PKU, a disorder in which children are unable to process certain proteins and in which poor metabolic control may result in neurocognitive deficits and behavioral problems, parents of children with PKU who reported an external attribution style (i.e., belief that their child's condition was outside of their control) had children who evidenced poorer metabolic control, a marker of treatment adherence, than children of parents who reported belief that health conditions were manageable (Antshel, Brewster & Waisbren, 2004). Thus, families' global beliefs about the controllability of health and illness appear related to the actions families take (e.g., engaging in prescribed treatment behaviors) to control and manage children's health.

These findings are similar to those reported in the pediatric obesity literature wherein parent locus of control predicted adherence to a group-based, empirically-supported behavior and nutrition intervention for childhood weight loss (Rosno, Steele, Johnston & Aylward, 2008). Parents who rated their child's weight problem as being due to chance (i.e., had external attributions) had children who attended fewer treatment sessions. Conversely, parents who indicated that the child was more responsible for his or her weight problem (i.e., had internal attributions) had children who had greater decreases in the number of unhealthy foods consumed at the end of treatment. Further, parents who report confidence in "powerful others" such as treatment providers had children who were more successful in treatment (Rosno et al., 2008). Taken together, these findings suggest that parents who place confidence in treatment providers and endorse a belief that individuals are in control of what happens to them may be more open to treatment recommendations compared to those who endorse beliefs that weight management is out of their control.

Thus, families adherence to prescribed medical treatment is influenced by a multitude of factors including understanding about the specific individual treatment regimen, the complexity of the treatments prescribed, families' beliefs about the importance of various elements of treatment, and families' beliefs about the amount of control they have over health outcomes. With this understanding of some factors that contribute to individuals' and families' likelihood to complete treatments as prescribed, we turn to an in depth look at the rationale behind one aspect of CF treatment: nutrition.

Impact of Nutrition on Pulmonary Function and Disease Progression

CF is a complex disorder involving multiple interrelated systems, and the specific contribution of each system to the presentation of CF is not fully understood. However, the association between nutrition and lung function has been long recognized (Bell, Durie & Forstner, 1984; Borowitz et al., 2002; Ramsey et al., 1992; Stallings et al., 2008).

Importance of Nutrition in Disease Management

The correlation between nutrition status and lung disease severity has been long known in CF research. Reviews of the U.S. CFF Patient Registry (CFFPR) demonstrate the strong positive relationship between nutrition status (measured by BMI percentile for children and BMI for adults) and lung function as measured by FEV₁ (CFF, 2007). Given these robust and consistent findings, it is recommended that all children with CF maintain weight above the 50th percentile (Stallings et al., 2008). However, despite better health outcomes associated with better nutrition, patients with CF are at increased risk for malnutrition because of the combination of increased metabolic demands, malabsorption of nutrients, and decreased intake (Bell et al., 1984; Lai & Farrell, 2008). Indeed, recent studies have found that 23% of children are below the 10th percentile weight for their age and sex (Stallings et al., 2008). Chronic malnutrition can result in

stunted growth, cognitive dysfunction, and increased susceptibility to lung disease (Lai & Farrell, 2008). Given the severity of this problem, current dietary recommendations include calorie consumption of 120% to 200% of the RDA for healthy individuals, with 40% of those calories coming from fat (MacDonald, Holden & Harris, 1991; Stallings et al., 2008).

Pathogenesis of Malnutrition

Malnutrition in CF stems from a combination of increased energy expenditures, increased energy loss, and reduced energy intake (Durie & Pencharz, 1989; Lai & Farrell, 2008). Research suggests that patients with CF have higher resting metabolic rates than what would be expected for age, size, and gender (Anthony et al., 1998; Buchdahl et al., 1988; Lai & Farrell, 2008; Tomezsko et al., 1994; Vaisman, Pencharz, Corey, Canny & Hann, 1987). This increased energy expenditure is hypothesized related to several factors, including increased work associated with breathing, cellular metabolism, and chronic infections (Lai & Farrell, 2008). Infections by specific bacterium have been shown to increase metabolic rate (Pencharz, Hill, Archibald, Levy & Newth, 1984), and there is evidence suggesting phenotypic differences associated with the basic genetic defect are related to differential rates of expenditure at the cellular level (Lai & Farrell, 2008). Although higher energy expenditures were associated with poorer lung function, increased metabolic rate has been observed in infants without advanced lung disease (Durie & Pencharz, 1989), demonstrating the importance of ensuring adequate nutrition early in a child's life.

In addition to elevated energy expenditures, CF patients are unable to absorb the nutrients that they take in. Obstructions in pancreatic ducts responsible for secreting digestive enzymes result in decreased capacity to digest and absorb fat and other essential nutrients (Lai & Farrell, 2008). Pancreatic insufficiency contributes to malabsorption of up to 40% of daily fat intake;

further, excessive mucus in the digestive tract may also limit absorption of other nutrients (Durie & Pencharz, 1989; Lai & Farrell, 2008). Thus, children with CF are unlikely to absorb all of the calories they consume due to a physiological inability to produce sufficient digestive enzymes.

To achieve energy balance, CF patients must take in significantly more calories than healthy peers (Stallings et al., 2008). However, research demonstrates that CF patients frequently fall below the RDA (Bell et al., 1984; Buchdal et al., 1988; Tomezsko, Stallings & Scanlin, 1992). Further, complications of CF including pain and vomiting associated with coughing, bloating and cramping, and restricted intake during pulmonary exacerbations can suppress appetite (Durie & Pencharz, 1989; Lai & Farrell, 2008). It is necessary for children to take in energy at the time when they feel least like eating to prevent further weakening and resultant lung damage.

When lung disease is mild or moderate and energy intake is adequate, it may be possible to achieve energy balance with increased intake. However, as pulmonary function declines and more energy is required to breathe, getting adequate energy from food becomes increasingly difficult and energy may be drawn from sources of energy within the body, eventually resulting in wasting of lean tissue (Schöni & Casaulta-Aebischer, 2000). If respiratory and skeletal muscle is lost, coughing and other processes required for airway clearance are compromised, leading to further infection and deterioration of lung function (Durie & Pencharz, 1989; Schöni & Casaulta-Aebischer, 2000). This cycle of energy imbalance and deterioration is thought to contribute to the rapid deterioration of pulmonary function and to eventual mortality.

Early Research on the Relation between Nutrition and Survival in CF

The correlation between lung function and nutrition has long been known (Bell et al., 1984; Chase, Long & Lavin, 1979; Lester et al., 1986; Ramsey et al., 1992; Roy, Darling &

Weber, 1984; Shepherd et al., 1986). Attention to the impact of nutrition on pulmonary function increased following the publication of a landmark study comparing CF patients from treatment centers in Boston and Toronto on growth and pulmonary indices. At the time of the comparison, the nutrition recommendations given to patients at the two centers were vastly different. Patients in Boston were instructed to eat a low-fat diet consistent with the recommendations to minimize possible discomfort associated with loss of fat via the stool (Bell et al., 1984). In contrast, patients in Toronto were instructed to eat an unrestricted fat diet and were encouraged to supplement intake with additional calories via fats. Both groups performed similarly on measures of pulmonary function; however, the median survival age for the Toronto patients was 30 years, compared to only 21 years for the Boston cohort (Corey, McLaughlin, Williams & Levison, 1988), suggesting that the higher calorie unrestricted fat diet conferred a survival advantage.

Given the overwhelming challenge presented by energy deficits, medical teams frequently seek to supplement caloric intake via oral, enteral (directly into the gut), or less frequently, parenteral (via vein) routes. A meta analysis evaluating the efficacy of various nutrition interventions in the treatment of under nutrition in CF has demonstrated very large effect sizes and clinically significant benefits for weight gain (Jelalian, Stark, Reynolds & Seifer, 1998). However, research examining the relation between nutritional status and longevity suggests that whereas surgical interventions produce short-term benefits, results are not long lasting. An examination of 10 undernourished children with CF who received aggressive enteral nutrition supplements compared to 14 age matched controls who received the usual care demonstrated effects on growth, cellular deterioration, and pulmonary function in the children who received supplemental nutrients. After at least one year of supplemental nutrition of approximately 120%-140% of RDA for protein and energy, the study group of malnourished

children demonstrated clinically and statistically significant changes in height and weight, with children achieving catch up growth. Children who received supplemental nutrition also experienced a statistically significant reversal in their declining pulmonary function and experienced fewer exacerbations compared to both the control group and their baseline level of functioning (Shepherd et al., 1986). After three years, the group who received supplemental nutrition achieved catch-up weight gain and their rate of decline in pulmonary function was significantly slower than the decline in the group who did not receive supplementary nutrition. However, after five years, these differences were no longer present, suggesting that improvements following limited, though intensive, nutrition therapy are short lived (Dalzell et al., 1992). Thus, surgical supplemental nutrition is effective in temporarily increasing weight and promoting catch-up growth in malnourished children. However, these changes begin to disappear following cessation of the intervention.

Recent “Causal” Research

The causal relationship between nutrition and pulmonary decline remains unclear, and given the complexity in the physiological interrelations among pulmonary, gastrointestinal, and endocrine systems, a simple and direct causal relationship is unlikely to emerge. However, population based research examining longitudinal data from large state- or nation-wide registries of CF patients has repeatedly demonstrated an important temporal relationship between nutrition and pulmonary function (Milla, 2007). This relationship suggests maintenance of nutrition status is an important determinant in the maintenance of lung function.

In a retrospective study of two birth cohorts, McPhail and colleagues (2008) compared lung function and markers of nutrition in 144 patients with CF divided into two groups based on year of birth. Children in the latter cohort had significant improvements in weight, height, BMI

percentile, and lung function compared to the children in the former cohort, and the children in the latter cohort experienced a significantly less steep rate of FEV₁ decline from the ages of 6-12 years than did the former cohort. This slowed rate of decline in pulmonary function was independently associated with higher BMI percentile and slower BMI percentile decline (McPhail, Acton, Fenchel, Amin & Seid, 2008). With all other factors held constant, rate of worsening of lung health was related to weight and ability to maintain weight. Though the comparison of birth cohorts presents a threat to the internal validity of the research in the form of possible historical or cohort effects, this study contributes to a body of research that consistently demonstrates relationships between weight and lung function.

In a prospective study examining the relation between children's rate of growth and pulmonary function, Thompson and colleagues found that children who demonstrated growth within an expected range experienced a much slower rate of decline in their pulmonary function than did children who did not demonstrate the expected growth (Thomson et al., 1995). Population analyses demonstrate similar results; in a report from a large German database that examined both cross sectional and longitudinal data, researchers found that malnourished patients with and without virulent pulmonary bacteria demonstrated significantly lower pulmonary function than well nourished patients with the same infection status. Further, longitudinal analysis demonstrated predictive relationships among weight and pulmonary function. During the one-year study period, children who were normally nourished and maintained or improved their weight for height maintained stable pulmonary function. Normally nourished children who lost 5% or more of their weight for height experienced an average decline of nearly 13% of predicted lung function. Children who were malnourished at the beginning of the study period continued to lose lung function with maintenance of weight for

height or loss of weight; however, those children who began the period malnourished and gained weight, gained approximately 4% of predicted lung function over the study period (Steinkamp & Wiedemann, 2002). Children's increase, maintenance, or loss of weight during the one-year study period was closely related to children's increase, maintenance, or loss of pulmonary function within the same period. Several key findings must be highlighted in this research. First, regardless of infection status, increased body weight was associated with better lung function. Second, the direction of the trend in children's weight mirrored the direction of change in children's lung function.

Similar associations are present in other evaluations of prospectively collected population data. An examination of data collected from clinics in Minnesota demonstrated the expected relationship between nutrition status and pulmonary function, but it also demonstrated an association between weight gain and improvements in pulmonary function. Children who consistently gained weight throughout the two-year study period experienced a significant gain in their FEV₁ during the study period. This gain in FEV₁ was not seen in children who lost weight one or more times during the study period. Further, these patterns did not vary by sex. The authors suggest this relation between improved pulmonary function and stable, consistent weight gain may be due to better lung growth in children whose bodies grew consistently (Peterson, Jacobs & Milla, 2003). This study suggests that malnourished children may experience improvements in lung function following weight gain. Given the permanent nature of lung tissue damage, any increase in pulmonary function is likely a reflection of improved airway clearance. Research examining the impact of early weight and nutrition status on later lung function suggests that a critical early window exists in which to achieve optimal pulmonary function.

There are a handful of studies that present compelling evidence demonstrating the relation between early weight status and subsequent pulmonary function. This body of research suggests that later pulmonary function is closely tied to early weight status, demonstrating a temporal relationship that is necessary but not sufficient for establishing a causal relationship. These findings highlight the critical importance for early intervention to improve nutrition status in patients with CF. An evaluation of 931 children in the US and Canada who were followed prospectively from the age of 3 to at least the age of 6 revealed significant associations between weight for age at age 3 and pulmonary function at age 6. Specifically, children with weight for age below the 5th percentile at age 3 had an average FEV₁ of 86% (\pm 20) predicted at age 6 compared to those children with weight for age above the 75th percentile at age 3 who had an average FEV₁ of 102% (\pm 18) predicted at age 6 (Konstan et al., 2003). Thus, children who weigh more than 75% of their same age peers at age 3 demonstrate normal and healthy lung function at age 6, whereas children who weigh less than 5% of their same age peers at age 3 demonstrate a diminished lung capacity and evidence of pulmonary disease at age 6. Given that 23% of children with CF are below the 10th percentile weight for age (Stallings et al., 2008), the problem of correcting early underweight poses critical and pressing problems for the field.

Lai and colleagues (2009) investigated the pulmonary outcomes in a cohort of newly diagnosed infants with pancreatic insufficiency who were enrolled in treatment designed to help them “catch up” to a weight level comparable to their birth weight z score by age two. Researchers examined pulmonary outcomes at age six for the group of children who achieved catch up weight at age two (responders) and those who did not (nonresponders). Similar to the findings reported by Konstan and colleagues (2003), the responders who caught up to their z score for body weight at age two had an average FEV₁ at 6 years old of 99.5% (\pm 13.9%)

compared to nonresponders, who had an average FEV₁ at 6 years of age of 88.3% (\pm 18.5%). Further, analyses revealed that the relationship between responder status and FEV₁ at age 6 was significant regardless of infection status, and the growth achieved between age 2 and age 6 was not associated with pulmonary functioning at age 6 (Lai et al., 2009). These findings add to the mounting evidence that weight status during the toddler and preschool years is critical in the development of healthy lungs that are able to withstand the assaults of CF.

Despite the research demonstrating a temporal relationship between weight status and pulmonary function in children with CF, the precise nature of underlying causal mechanisms is still unknown (Schöni & Casaulta-Aebischer, 2000). However, research examining pulmonary development in nutritionally deprived rats has provided some insight into potential mechanisms affecting pulmonary development in humans. Research into the effects of nutritional deprivation in newborn rats has demonstrated both immediate and long-term effects of malnutrition on the physiological composition of the lungs, including changes in the size of the alveoli and in the development of elastic lung tissue. Further, these changes persist after adequate nutrition is restored (Milla, 2007). The long-term impact of inadequate development of elastic lung tissue is thought to be related to air-trapping that occurs in the lungs of infants with CF and that is correlated with malnourishment (Milla, 2007). Unlike rats, however, there is evidence that pulmonary effects from low birth weight in human infants can be corrected. Lai and colleagues (2009) investigation suggests that children who demonstrate catch up growth before the age of 2 have fewer symptoms and better pulmonary function at age 6 than children who were not able to regain their expected weight (Lai et al., 2009). Thus, animal models provide insights into potential mechanisms underlying the relation between weight and pulmonary function and underscore the importance of early intervention to achieve and maintain optimal lung health.

Children with CF face an uphill battle in their struggle to maintain adequate weight. Children with CF require more energy to breathe, absorb energy more poorly than children without CF, and often have decreased appetites, yet the long term health and survival of children with CF is closely tied to children's ability to gain and maintain weight. Given the close relationship between weight and survival, current recommendations for children with CF range from 120% - 200% of RDA (Borowitz et al., 2002; Stallings et al., 2008). However, despite directions from medical providers, children's dietary intake typically falls far short of recommendations (Bell et al., 1984; Crist et al., 1994; Powers et al., 2002; Stark et al., 1995). This shortcoming leaves children vulnerable to a vicious downward spiral of energy imbalance exacerbated by symptomatic lung disease, decreased airway clearance, infection, inflammation, lung damage, and precipitous declines in health. Early intervention is critical to preventing or delaying this cycle and giving children the best opportunity to live full, long lives with minimal impact from their disease.

Given the importance in maintaining adequate nutrition status for the preservation of long-term lung health in children with CF, it is not surprising that a number of interventions exist to supplement caloric deficiencies. Jelalian and colleagues (1998) conducted a meta-analysis of nutrition intervention studies for weight gain in children with CF. Four classes of nutrition interventions were included in the analysis: oral supplements, enteral nutrition (EN; supplements delivered directly into the digestive tract via tubes placed in the nose or surgically implanted into the stomach), parenteral nutrition (PN; nutrition delivered directly into the bloodstream), and behavioral interventions. The present focus will be on oral, EN, and PN, as behavioral interventions will be discussed in depth later in this review.

Oral supplements and EN can be administered on an outpatient basis, but PN is typically delivered inpatient because of the risk of infection. All of the studies reviewed by Jelalian and colleagues (1998) used weight gain as an outcome measure, and several included caloric intake as outcome measures as well. The studies included in the analysis included six with oral supplementation ranging in treatment duration from 3 to 30 months, five with EN ranging in duration from 12 days to 24 weeks, and three with PN ranging from 9 to 30 days. Weighted effect sizes were calculated to facilitate comparisons among treatments, and effect sizes were in the very large range (Cohen, 1977), with 1.62 for oral supplements, 1.78 for EN, and 2.20 for PN (Jelalian et al., 1998), demonstrating efficacy for all three nutrition interventions for weight gain. However, long-term weight gain has been demonstrably difficult to maintain using only oral supplements or surgical interventions (Shepherd et al., 1986; Stark, Bowen, Tyc, Evans & Passero, 1990). Given the lack of long-term efficacy of surgical interventions at promoting weight gain, anecdotal reports from parents about oppositional behavior (Modi & Quittner, 2006) mealtime conflicts following parents' efforts to increase children's intake at family meals, and efficacy of behavioral interventions in pediatric feeding disorders (Linscheid, Budd & Rasnake, 2003), researchers began to investigate the mealtime behavior of children with CF explore the relation between children's behavior at mealtime and dietary intake.

Mealtime Family Interactions and Cystic Fibrosis

Given the long recognized association between nutrition and health in CF, treatments to improve weight have long been part of CF treatment, including surgical supplementation (Bowen & Stark, 1991; Jelalian et al., 1998; Stark et al., 1990). Although these interventions were generally successful in increasing weight during the intervention phases, weight and intake typically return to low baseline levels following intervention (Dalzell et al., 1992; Jelalian et al.,

1998; Shepherd et al., 1986; Stark et al., 1990). Given successes in behavioral interventions for feeding disorders for children with failure to thrive (Babbitt et al., 1994), food refusal (Babbitt et al., 1994; Cooper et al., 1999; Singer, Ambuel, Wade & Jaffe, 1992; Wolff & Lierman, 1994), and in children who developed feeding problems secondary to chronic illness or congenital anomalies (Handen, Mandell & Russo, 1986; Palmer, Thompson & Linscheid, 1975) and parents' anecdotal reports of mealtime conflicts resulting from attempts to encourage eating (Singer, Nofer, Benson-Szekely & Brooks, 1991; Stark et al., 1993), several researchers began to investigate the incidence of behavioral problems at mealtime in children with CF to better understand the problem of non-eating.

Research examining behavior during meals in families of children with CF in comparison to healthy controls (Mitchell, Powers, Byars, Dickstein & Stark, 2004; Powers et al., 2002; Powers, Mitchell et al., 2005; Stark et al., 1995, 1997) and to children with feeding problems (Sanders, Patel, Le Grice & Shepherd, 1993; Sanders, Turner, Wall, Waugh & Tully, 1997) has uncovered consistent patterns of mealtime behavior related both to the stress reported by parents and to children's difficulty gaining weight. Researchers offered a behavioral conceptualization to explain the relation between parental encouragement and children's non-eating behavior (Bowen & Stark, 1991). Briefly, parents experience intrinsic pressure and pressure from treatment teams to increase children's weight and intake; this leads to an increased focus on mealtime behavior and increased attention to any behavior inconsistent with parental goals to increase weight. Parents attend disproportionately to children's not eating, and this increased attention inadvertently rewards non-eating further reducing children's intake by increasing behaviors incompatible with eating (Bowen & Stark, 1991; Crist et al., 1994; Singer et al., 1991; Stark et al., 1995). Thus, parental attempts to encourage children's compliance to dietary

recommendations often backfires and serves to increase children's oppositional behavior and reduce children's consumption.

Parent reports of challenging behavior

Researchers asked parents to report on their children's behavior at mealtimes to better understand factors that contribute to non-eating. Research examining parental stress and parent and child mealtime behavior in families with and without CF revealed interesting associations (Crist et al., 1994). Parents of 22 children with CF and a matched control group between one and 8 years of age, completed diet diaries, the Behavioral Pediatric Feeding Assessment Scale (BPFAS), a measure of parent and child behaviors associated with poor nutritional intake, and the Parenting Stress Index (PSI), a measure of stress within the parent-child system. Overall, parents of children with CF reported more mealtime behaviors incompatible with eating and a greater number of problems at mealtime than did parents of healthy children. Specifically, significantly more parents of children with CF endorsed problems such as "has problem chewing food" (33%), "chokes or gags at meal" (33%), "takes longer than 20 minutes to finish meal" (55%), "has poor appetite" (48%), "reluctant to come to mealtime" (43%), "spits out food" (41%), and "delays eating by talking" (46%) than did parents of control children. Further, parents of children with CF endorsed using problematic strategies to get their child to eat, including coaxing (68%), making a second meal if the child refused the first one (55%), getting angry (27%), making threats (23%), and force feeding (14%). Parents of children with CF endorsed the belief that their child's eating hurts his/her health significantly more than did parents of control children. These findings fit perfectly within the behavioral conceptualization of mealtime conflicts. Parents endorsed the belief that children were harmed by their (not) eating

and engaged in strategies that reinforce noncompliance (e.g., coaxing or making a second meal), and children engaged in more problematic behaviors.

Further, parents of children with CF reported significantly more child behaviors as a cause of stress on the PSI than did parents of control children. In addition, there was a significant negative relationship between caloric intake and parent report of both parent and child eating-incompatible behaviors as well as a significant negative correlation between caloric intake and the child behavior domain of the PSI, so that increased parent report of problematic behavior was associated with reduced caloric intake (Crist et al., 1994). Thus, parents of children with CF reported that child behaviors were a cause of parental stress and also reported higher levels of behavior that are incompatible with eating. This finding provides support for Bowen and Stark's (1990) behavioral conceptualization that posits increased parental stress contributes to parental mismanagement, which may reinforce non-eating, decrease children's rate of eating, and limit exposure to a wide variety of food. However, this cross sectional analysis leaves open many questions about the functional relationship among these behaviors.

Powers and colleagues (2002) compared 35 infants and toddlers with CF (mean age 18.6 months, SD = 8.1 months) to matched controls on a variety of anthropometric measures and the BPFAS. Parents of children with CF identified significantly more child behaviors as problematic than did parents of control children. The most frequently cited problem behaviors by CF parents were problems with their child's willingness to try novel foods (48%) and eat vegetables (48%). Parents of children with CF also reported significantly more distress than did control parents; more than one third of parents of children with CF (37%) reported feeling anxious or frustrated when feeding their children, and one third of parents reported belief that their child does not eat enough (32%). Notably, parents of children with CF reported significantly higher use of

ineffective parenting strategies than did parents of control children, including coaxing (26%), to increase their child's intake. Correlation analyses suggested that children's meal duration was positively associated with parent report of problematic child behaviors; however, no relationship emerged between child behavior and calories consumed. Meal duration in children with CF was found to be significantly longer than healthy controls, consistent with findings from other age groups (Stark et al., 1995, 1997). However, despite the increased duration of meals for children with CF, these children were not found to consume more during the meal or spend a higher percentage of time eating (Powers et al., 2002), suggesting that increasing meal duration is an ineffective strategy for increasing intake. These findings indicate that parents of children with CF experience more affective distress at mealtimes, see more child behaviors as problematic, and engage in more ineffective strategies than parents of healthy children.

Observational reports of child eating behavior

Observational research examining patterns of eating has sought to uncover differences in intake between children with and without CF. However, analysis of videotaped meals suggests that some behavior is consistent regardless of disease status. For example, toddlers and preschoolers with CF eat a similar diet at the same rate as those without CF when expressed in both bites and sips per minute and in 10-second intervals containing bites (Powers et al., 2002; Powers, Mitchell et al., 2005; Stark et al., 1995). Further, each bite contained approximately the same number of calories (Stark et al., 1995), so children with CF were not taking smaller bites than healthy control children. However, significant differences in meal duration and calories per meal were noted, with children with CF taking longer to eat meals and consuming a greater number of calories during the dinner meal (Stark et al., 1995), suggesting that the extra calories toddlers consumed were taken in at the end of the meal. This finding is in contrast to Powers and

colleagues (2002) findings that increased meal duration was not associated with increased consumption. These discrepant findings suggest that extending meal duration may be an inconsistently effective short-term strategy for increasing intake, thus reinforcing parents' use of that strategy, but that the strategy itself may not be consistently effective in the long term.

Despite the increased caloric intake, children with CF were at a lower percentage of ideal body weight than the control children (Stark et al., 1995). These findings were replicated in a sample of school-age children with CF (Stark et al., 1997). Overall, children with and without CF were not behaving very differently; the two groups took approximately the same number of bites at the same rate of speed, but mealtimes for CF children were longer. Despite these longer mealtimes, children with CF were still underweight.

Research exploring eating behavior in infants and toddlers lends additional support to the finding that children with CF initially exhibit similar eating behavior to children without CF in terms of pace of eating, but despite these similarities, parents of children with CF report higher levels of problem behavior and many report rates of anxiety or frustration when feeding their children. Powers and colleagues (2002) compared 35 infants and toddlers (mean age 18.6 months, SD = 8.1 months) and matched controls on measures of height, weight, weight percentile for age, weight-for-height percentile, and z scores for height-for-age, weight-for-age, and weight-for-height; analyses revealed only one statistically significant difference between the groups on z score height for age, with the control group being taller than the CF group. Powers and colleagues found deficits in caloric intake for the CF sample, with only 11% meeting the CF dietary recommendation. Further, the groups did not differ on comparisons of average daily caloric intake, but they did differ on calories consumed per kilogram of weight, with CF children consuming more calories per kilogram than healthy controls.

This study by Powers and colleagues supported Stark and colleagues' (1995, 1997) findings that children with CF typically eat the same amount and at the same rate of speed as do children without CF. Further, Powers and colleagues found that infants and toddlers with and without CF consumed equivalent calories per meal. However, children with CF took longer to eat their meals than did healthy controls, and within the CF group, meal duration was negatively correlated with weight-for-age, so that lighter children took longer to eat than heavier children. Similarly, a positive relation between weight percentile for age and amount of calories per bite emerged. Together, these two findings may reflect either larger bites or higher calorie food presentation for children with a higher weight percentile for age (Powers et al., 2002), indicating that minor changes in meal preparation through the addition of calorie-dense foods could improve overall intake.

Parent and Child Behaviors at Mealtime

Observational research exploring parents' and children's behaviors at mealtime lend support to Bowen and Stark's (1991) conceptualization of an interactive pattern of parent and child behaviors at mealtime that serve to reinforce children's non-eating. Sanders and colleagues (1997) videotaped and coded the meals of 25 children with CF, 25 children with feeding problems, and 20 healthy controls. All participants were between the ages of 12 and 84 months and were matched to the CF group for age, gender, and family SES. The coding system used by Sanders and colleagues (1997) allowed for the identification of child appropriate (e.g., requesting food, taking a bite, verbal interactions) and disruptive (e.g. food refusal, play with food, leaving the table) behaviors as well as parent aversive (e.g., prompt, eating comment, social instruction in response to disruptive behavior) and non-aversive (e.g., praise, prompt, specific instruction, social attention in response to appropriate behavior) behaviors. Researchers also assessed

calories consumed at the target meal, parenting sense of competence, and children's anthropometrics.

Sanders and colleagues (1997) found no difference between the CF and healthy control groups in weight for age and overall mother positive behavior, but found that mothers of CF children engaged in more aversive behavior during the meal and provided more specific instructions than mothers of healthy controls. Further, mothers of children with CF provided more prompts (both aversive and non-aversive) than mothers of healthy children and children with feeding problems. Further, CF children demonstrated more noncompliance and had fewer appropriate verbal interactions than healthy children; however, CF children played with their food less than children with feeding problems and as much as healthy children. Both parents of CF children and children with feeding problems reported more difficulty at mealtime than parents of healthy children, and parents of CF children reported that the mealtime behaviors are more problematic than did parents of healthy children. Further, parents of CF children reported a lower sense of self-competence in parenting than did parents of children with feeding problems or healthy children. Overall, mothers of children with CF displayed higher levels of aversive behavior, lower levels of positive attention, more prompts, and more instructions than did other mothers in the study. Researchers suggest that this high level of involvement and instruction by parents of children with CF increases the opportunity for children's noncompliance and reduces the likelihood that children will learn to eat independently without parental involvement (Sanders et al., 1997). Children with CF experienced more aversive behavior from their mothers and engaged in fewer appropriate verbal exchanges with family members. Parents of children with CF had a low sense of self-confidence about their ability to parent, reported high levels of stress

about children's eating, and engaged in high levels of aversive strategies to encourage eating. Taken together, mealtimes appear to be stressful and aversive for both children and parents.

Powers, Mitchell and colleagues (2005) found similar patterns of behavior among families of children with CF aged 7 to 35 months when compared to age, gender, and SES-matched healthy control families. Researchers used the Dyadic Interaction Nomenclature for Eating (DINE) developed by Stark and colleagues (2000) to code dinners recorded at the family home. The DINE captures information about eating behavior (e.g., frequency of bites and sips) as well as parent and child interactive behaviors (e.g., parent commands and coaxing, and child leaving the table or refusing to eat). Researchers found that CF parents gave their children more direct commands and fed their children more than parents of healthy children. They also found that children with CF took more frequent bites than did healthy controls. In both groups, shorter meals were associated with more frequent bites, and children in both groups increased their rates of food refusal and walking away from the table during the second half of the meal (Powers, Mitchell, et al., 2005). These findings underscore the similarity in behavior between CF and healthy toddlers and provide support for the theory that maladaptive parent behavior (e.g., direct commands or coaxing) increases opportunity for child noncompliance and contributes to high-conflict mealtimes. In addition, these findings provide insight into reasons why parent aversive behavior, despite its unpleasant nature, persists in families of children with CF. Children of CF took more frequent bites than healthy controls, suggesting that parents' strategies may be immediately reinforcing (i.e., following a command, children take a bite). Further, given reports of parental stress in other studies (Crist et al., 1994), children's eating may also serve to temporarily decrease parental anxiety. However the long term effect of this interaction of

parental feeding strategies and child behavior appears to be a reduction in the overall quality of family interaction.

Indeed, researchers coding mealtimes from the sample described above (Powers, Mitchell et al., 2005) using a coding system that measures the quality of family functioning on domains such as communication (i.e., exchange of ideas and information), interpersonal involvement (i.e., demonstrating respect for and interest in each other), affect management (i.e., relevance and appropriateness of expressed emotion), behavior control (i.e., maintenance of behavior standards), roles (i.e., establish and fulfill family functions), and a composite index of overall functioning, uncovered relations among quality of family functioning and child feeding behavior. Researchers compared families of infants and toddlers with CF and age, gender, and SES-matched peers (Mitchell et al., 2004), and differences between the groups of families emerged. Specifically, significantly more CF families fell into the “unhealthy” range for overall family functioning, communication, affect management, and interpersonal involvement than did families of healthy children. Further, those families of children with CF who demonstrated fewer problems were those who reported lower rates of disruptive parent management behaviors and lower problem ratings on the BPFAS (Mitchell et al., 2004). Thus, families who reported fewer aversive parent behaviors and rated all behaviors as less problematic were less likely to demonstrate maladaptive patterns of family functioning.

In sum, children with CF are at risk for malnutrition due to a combination of factors that create an energy imbalance; however, malnutrition is strongly associated with decline in pulmonary functioning, longer recovery following exacerbations, and poorer survival (Konstan et al., 2003; Lai et al., 2009; Roy et al., 1984; Steinkamp & Wiedemann, 2002). Surgical interventions to address malnutrition provide short-term benefits but do not result in long-term

change (Jelalian et al., 1998; Shepherd et al., 1986). However, observations of CF families of toddlers, preschoolers, and school-age children at mealtime reveal consistent patterns of parent and child behavior that inadvertently reinforce non-eating and may contribute to deterioration in family functioning. Further, parents consistently demonstrate their awareness regarding the importance of intake in maintaining lung health by placing a high priority on children's intake; however, the problem appears to be parents' strategies. Behavioral theory states that when a behavior is followed by a desirable outcome, that behavior is likely to be repeated. Parents of children with CF consistently attend to children's non-eating (Powers, Mitchell, et al., 2005; Sanders et al., 1997; Stark, 2003b), and as parental attention is rewarding for children, children with CF are likely to continue behavior incompatible with eating. Similarly, those occasions in which parent prompting precedes a child bite serve to reinforce parental use of these strategies. Unlike in healthy children whose parents may be able to rationalize worries about not eating, the pressure of increasing intake is a constant one for families, "for the parent of a child with CF one more bite is always better and one step closer to optimum health" (p. 795, Stark, 2003b). Given the behavioral basis of parents' deficits, early intervention to provide parents with the skills to promote appropriate eating behavior may prevent malnutrition, decrease familial stress, and slow the decline of pulmonary function.

Behavioral Intervention to Improve Intake in Children with CF

Following the identification of consistent patterns of behaviors incompatible with eating in children and families with CF, interventions to address these patterns of mealtime behaviors were developed. In 1990, Stark, Bowen and colleagues piloted a behavioral intervention to increase calorie consumption for five mildly malnourished school-age children with CF whose health status did not warrant inpatient surgical nutrition intervention (Stark et al., 1990). This

intervention combined nutrition education and parent training in a six-session group treatment format that increased calories sequentially across meals and taught parents behavioral strategies to motivate children to consume more calories. The basic format of this treatment (Stark, 2003a) has served as the template for the majority of the behavioral nutrition interventions for children with CF (Stark, 2003b; Stark et al., 1993, 1996, 2009; Stark, Opiari, et al., 2003; Stark, Powers, Jelalian, Rape & Miller, 1994; Powers et al., 2003; Powers, Jones et al., 2005). The original intervention utilized a group format and convened both parent and child groups, with the child group learning age appropriate calorie and nutrition information and earning stickers on a reward tracking chart for meeting calorie goals for meals each week. Parents met with a clinical psychologist and a pediatric dietitian for 90 minutes each session while children met simultaneously with a psychology graduate student and undergraduate research assistant. Individualized calorie goals were developed using information from the baseline food diaries and targeting total caloric intake goals at 130% of the RDA for healthy children. The first session focused exclusively on training parents and children to monitor and record caloric intake; the second through fifth sessions focused on increasing calories in snacks, breakfast, lunch, and dinner, and introduced parents to behavioral methods to motivate their children to increase food consumption. Sessions two through five focused on describing and praising appropriate child behavior, planned ignoring of child complaints, structuring mealtimes, and implementing home-based contingencies for children's making calorie goals. The final session focused on fading intervention-based contingencies and planning for maintaining calorie goals during sick days.

Effectiveness in Promoting Behavior Change in Malnourished Children

In Stark's original pilot study (Stark et al., 1990), five children, ages 5 years 10 months to 12 years 1 month whose weight for age ranged from below the 5th to the 35th percentiles and their

parents participated. Dependent measures included caloric intake, child weight and body fat, and pulmonary function tests. Families kept food diaries prior to the intervention, throughout the intervention, and at one month and three month follow up. Anthropometric and pulmonary function measurements were taken before, during, and after the intervention. Over the course of the intervention, children's caloric intake increased an average of 1,050 calories per day (51% increase over baseline) and was maintained at 9-month follow up. Evidence for the effectiveness of this single case intervention can be understood through a visual inspection of the graphs plotting intake at each meal by the weeks of the intervention. Caloric intake for each meal increased following the session targeting that meal and remained at elevated levels for the remainder of treatment. Children also showed significant increases in rate of weight gain, weight percentile, and body fat percentage from pre- to post-treatment. Further, these changes appeared to be long lasting; in the year after treatment, children's rate of weight gain was more similar to that of healthy children their age than in the year prior to treatment (Stark et al., 1990). This first study provided support for a combined nutrition and behavior parent and child group intervention to improve nutrition status in underweight school-age children with CF.

One replication of Stark and colleagues' (1990) original study incorporates observations of mealtime behavior to measure distribution of bites throughout the meal and pace of eating (Stark et al., 1993). As with the first study, the three children in the replication increased their total daily caloric intake from baseline levels. Children's baseline consumption as expressed in %RDA improved from 94%, 99% and 114% to 116%, 152% and 175%, respectively. As children are expected to continuously gain weight, children's velocity of weight gain served as a marker of treatment success. Children improved their rate of weight gain in the year following the intervention compared to the year preceding the intervention. Analysis of children's

mealtime behavior indicates that children increased their calories consumed per minute from pre-treatment to 48-week follow up. One child increased her caloric intake with no change to meal duration, another child increased her caloric intake and decreased meal duration, and the third child nearly quadrupled his caloric intake while increasing the duration of his meal from 20 to 30 minutes (Stark et al., 1993). This study provides evidence for the success of a behavioral intervention in increasing school-age children's consumption as well as the long-term maintenance of treatment effects.

Effectiveness in Promoting Behavior Change in Adequately Nourished Children

Problematic mealtime behaviors are not unique to families with underweight children (Mitchell et al., 2004; Powers et al., 2002, Powers, Mitchell, et al., 2005; Sanders et al., 1997; Stark et al., 1995), and research has demonstrated that the behavioral interventions effective in increasing intake and decreasing mealtime conflict can improve parent-child interactions at mealtime for weight-appropriate children with CF (Stark et al., 1994). Families of a two-year old boy in the 90th percentile of weight for age and a five-year old boy at the 55th percentile of weight for age enrolled in an individualized version of Stark's original group treatment. Mealtime behaviors were filmed and diet diaries were collected throughout the intervention. The 5 year-old boy increased his weight percentile from 55th to 78th during the intervention and to the 93rd at 8 month follow up. The 2 year-old boy's diet became more appropriate for his age in that it dramatically increased the proportion of calories consumed via solid foods. He did not increase overall caloric intake, but he gained weight during the intervention and his weight percentile remained relatively stable throughout the study period, remaining at 90th percentile at post-treatment and 88th at one month and one year follow up appointments. Changes in behavior during mealtimes filmed at home indicate that parents were implementing the strategies taught

during the intervention. Parents reduced their attention to child disruptive behavior, increased attention to appropriate eating, and improved contingency setting (i.e., limiting length of mealtime). Further, child appropriate behavior increased and disruptive behavior decreased over the course of the intervention (Stark et al., 1994), suggesting that behavioral parent training targeted specifically for mealtimes is effective in changing parent and child behavior in families of adequately nourished children with CF as well as further improving the nutrition status of toddler and preschool age children who are already above the 50th percentile.

Developmental Adaptations to Promote Behavior Change in Young Children

Given the importance of early weight and nutrition status in long-term pulmonary function and health outcomes (Konstan et al., 2003; Lai et al., 2009), effective early intervention is critical to achieving optimal lung health. Toddlerhood is an ideal age for early intervention because it is a developmental period where children begin to assume control of eating as they assert some autonomy, and behavioral feeding problems are likely to emerge and challenge parents (Linscheid & Rasnake, 2001; Satter, 2000). Indeed, observational research has identified that toddlers regardless of disease status display a higher frequency of challenging behaviors during meals than do school age children (Powers et al., 2002). Thus, the preschool age appears an ideal age at which to intervene to increase caloric consumption for maximum health benefit and before stable patterns of maladaptive behavior are developed.

Research demonstrates effectiveness of behavioral interventions with young children with severe underweight. Results from a non-manualized behavioral intervention for toddler and preschool children lend further to support to the efficacy of behavioral interventions to correct malnutrition in young children with CF (Singer et al., 1991). Four children ranging in age from 10 to 40 months who were diagnosed with CF and non-organic failure to thrive, were below the

fifth percentile weight for height, and consumed between 48% and 62% of the RDA were observed interacting with their parents during meals. Treatment consisted of developing a hierarchy of steps designed to reinforce eating and drinking using shaping, positive reinforcement, time-out, and ignoring. Treatment was initially conducted by treatment center staff and was gradually transferred to parents after children demonstrated consistent eating behavior. Following treatment, children's mean RDA improved from 54% to 92%, and three of the four children demonstrated intake at or above 100% of RDA. The one child whose intake did not improve to 100% prematurely terminated from the program, lived at a significant distance from the treatment facility, and reported family disruption and chronic psychological stressors. The other three children demonstrated catch up growth with percentiles weight for height ranging from the tenth to the fiftieth at follow-up appointments ranging from 7 to 24 months post-discharge (Singer et al., 1991). Thus, individualized, intensive, inpatient behavioral interventions have demonstrated success changing eating behavior and concurrently increasing weight percentiles of toddlers and preschoolers with CF.

Behavioral interventions appear to be effective with severely malnourished toddlers and preschoolers as well as with less severely malnourished children. Powers and colleagues modified Stark's intervention (Stark, 2003a) and have demonstrated success in changing parent and child behavior and improving nutrition status in toddlers and preschoolers. Powers and colleagues' (2003) intervention compared the effectiveness of a nutrition education intervention to a nutrition education plus behavior training intervention to eight families of toddlers and preschoolers with CF. Unlike earlier interventions tested with school age children which held eight treatment sessions across nine weeks, the eight sessions of Powers and colleagues' (2003) interventions spanned one year. The first four 60-minute sessions in both arms of Powers and

colleagues' study occurred over three months, and the final four sessions occurred approximately every other month. All families learned about the physiology of CF, dietary recommendations for energy and micronutrient intake, and enzyme therapy, and all families were taught a range of dietary strategies to increase energy intake. Those families in the nutrition plus behavior intervention also received behavior management training focusing on direct commands, differential attention, and time-out procedures using modeling and role-playing strategies both with the psychologist and with their child in session four. In both conditions, children's average intake increased from approximately 78% RDA to over 100% RDA. Children in the nutrition plus behavior intervention increased their average daily intake 406 calories per day (40% increase) and children in the nutrition-only intervention increased daily intake 285 calories per day (28% increase); these differences approached, but failed to reach statistical significance. The effect size for calories per day for the combined intervention was 2.23 versus 1.95 for the nutrition-only intervention – both large effect sizes (Powers et al., 2003). They found attrition to be a significant problem for the longer intervention and recommend a shorter, more intensive intervention with the possibility of booster sessions to enhance maintenance of treatment gains in the months post-treatment.

In a follow-up study, Powers, Jones, and colleagues (2005) compared nutrition-only to behavioral intervention for children aged 18 to 48 months using protocol similar to that used in their previous study (Powers et al., 2003) but with the timing shortened to occur over an 8-week period rather than one year. The timing of the intervention tested in this study more closely approximates the interventions tested by Stark and colleagues (Stark, 2003a, b; Stark et al., 1993, 1994, 1996, 2009; Stark, Opiari et al., 2003) and the timing proposed in the current study. Powers and colleagues used a crossover design so that families who initially participated in the

nutrition-only intervention had an opportunity to enroll in the combined intervention. Children in the combined intervention achieved a statistically significant increase in caloric intake compared to children in the nutrition-only intervention (+843 v. -131 calories per day) equivalent to a 60% increase in total calories. Children in the combined intervention also achieved the goal of obtaining 35% to 40% of calories from fat, and whereas none of the children in either group were consuming the recommended 120% RDA at pre-treatment, 100% (4 of 4) of children in the combined intervention met this goal at post compared to only 1 out of 6 in the nutrition only intervention. Further, these increases were largely maintained at 3- and 12-month follow-up, with all participants who received the combined intervention achieving 120% RDA at the 12-month follow-up. Children who participated in the combined intervention also achieved remarkable catch-up growth; seven of the 9 children who received the combined intervention achieved weight velocity equivalent to a child growing at the 50th percentile at the 3- and 12-month follow up. The two children who did not achieve the weight velocity benchmark were within 0.1kg (out of 1.0kg) of the expected gain at the 3-month follow up and within 0.3 and 0.6kg of expected 2.0kg growth at the 12-month follow up. All children achieved the benchmark for increase in height (Powers, Jones, et al., 2005).

These outcomes demonstrate considerable improvements in energy intake and growth for children who received the combined intervention and provide evidence of efficacy for behavioral intervention in improving children's nutrition status. Given the link between nutrition status at ages 2 and 3 and pulmonary function at age 6 (Konstan et al., 2003; Lai et al., 2009), this eight week combined behavioral and nutrition intervention has the potential provide enormous and long-lasting benefits to children with CF by improving long-term lung health and giving children the best opportunity to live full, long lives with minimal impact from their disease.

Outstanding Questions

These interventions demonstrate remarkable effects for children's caloric intake and growth and demonstrate the importance of combining nutrition education and behavior training for parents of toddlers and preschoolers with CF. However, research examining the adaptation of evidence based treatments (EBTs) for childhood mental health problems developed in university research labs to community mental health settings indicates there is much work to be done before these combined behavior and nutrition interventions can be more broadly disseminated (Southam-Gerow, Hourigan & Allin, 2009). Researchers interested in disseminating EBTs to children receiving care in community settings face a number of important steps between treatment development within a research context and widespread adoption within community practice settings. The previously described research by Stark, Powers, and colleagues establishes positive treatment outcomes in controlled research settings (i.e., *efficacy* research). However, additional research is needed to evaluate combined behavior and nutrition treatments for parents of toddlers and preschool children in a new, less controlled community service setting (i.e., *effectiveness* research) prior to transportability, implementation, and dissemination phases of research. It is necessary to determine whether previous findings can be replicated in a different context or with non-doctoral level providers.

Further, the hypothesized mechanism of change in this study is the behavioral intervention, but this body of research lacks systematic measurement of behavioral change. The question remains as to *what* behaviors changed in the families of these children. *How* did these changes come about? This question is important as it may provide guidance for improving treatment effectiveness. For example, if the primary skill used by parents was differential reinforcement of other behavior (DRO), and parents' use of DRO produced the majority of

change in children's mealtime behavior, then researchers should direct their attention toward the development of a highly efficient abbreviated DRO-specific intervention that can be delivered by a variety of professionals working in the CF clinics around the country (e.g., nutritionists, social workers, nurses) with the goal of reaching all parents of toddlers to maximize the opportunity for the greatest benefit. As another example, suppose that in contrast to the previous theory about a single critical technique (e.g., DRO), it is important for parents to develop skills in the complete repertoire of behavioral management techniques, and suppose again that most families evidence only minor skills deficits that require identification and brief intervention. If research examining changes in family behavior was able to identify markers of these deficits (i.e., on paper and pencil measures such as the BPFAS) linking the skill deficit to change in behavior caused by the intervention (e.g., a deficit in contingency management that improves following the session that addresses contingency management), then researchers are faced with a different treatment-development and dissemination imperative.

Application of Single Case Methodology

The single case methodology is well suited to these questions of whether changes occur as a function of the intervention. When carried out with methodological rigor, the single case design can demonstrate causal relations and rule out threats to validity and as such, should be considered an experimental approach. Single case designs differ from group designs wherein conclusions about the effectiveness of an intervention are drawn from the differences demonstrated between randomly assigned groups. The methodologically rigorous single case experiment involves four key characteristics that permit one to draw inferences about the effects of an intervention: continuous assessment, baseline assessment, stability of performance, and use of different phases (Kazdin, 2003). Each of the four will be reviewed briefly below before

turning to a discussion of why single case methodology is ideally suited to the proposed research question.

Continuous assessment. The application of continuous assessment is one of the primary differences between single case and between group methods. In a single case design, repeated observations of the target behavior or behaviors over time, including in the baseline phase and during the intervention, allow for the examination of the pattern, stability, and change in performance relative to the timing of the intervention. Unlike in group designs where conclusions are drawn about effectiveness by examining changes in group mean scores taken at relatively few assessment points (e.g., pre and post treatment), conclusions about the effects of the intervention for single case experiments are drawn by examining the pattern of many repeated assessments for a small number of subjects (Kazdin, 2003). These repeated assessments provide a clear picture of the behavior in question and occur at sufficient frequency as to identify the timing of changes. As Linscheid states, “measures must be capable of revealing not only the degree of change, but observations must be made frequently enough to reveal when change occurred,” (2000, pp. 438).

Baseline assessment. Critical to the understanding of the effect of an interaction is an understanding of the target behavior prior to the intervention. The data collected in the baseline (i.e., pre-intervention) phase of the experiment serves two purposes: data describe the current state of the target behavior (i.e., descriptive function) and provide the basis for predicting future behavior in the absence of intervention (i.e., predictive function) to evaluate whether the intervention leads to change (Kazdin, 2003). Ideally, the data collected in the baseline phase will demonstrate a stable pattern without much variability or slope. These issues will be discussed below.

Stability of performance. There are two characteristics of data collected during continuous assessment that require attention. The first is the variability in the data, or how much the assessment points fluctuate. Too much variability in baseline data makes predicting future behavior difficult. The second characteristic is the slope or trend in the data, or the extent to which the data points appear to be changing in a predictable way. Data trending in the direction of the intervention or highly variable data points during the baseline phase interfere with the ability to draw inferences about the effects of the intervention (Kazdin, 2003). Stable performance during the baseline phase is ideal, but following the introduction of the intervention, the researcher hopes for immediate and dramatic changes in target behavior that make attributions to chance variables logically implausible.

Use of different phases. The final key characteristic of a single case experiment is the use of different phases of the intervention. There are a variety of methods by which to vary the phases in an intervention, but the purpose of these phases is to determine whether the pattern of performance changes in the direction predicted by the intervention. For example, in an ABAB design, the baseline phase (A) and the intervention (B) are alternated. If the intervention is effective, it would be expected that the pattern of results demonstrated in the first baseline phase would return to that level during the second baseline phase, then change again at the onset of the second phase of the intervention. In the application of interventions where the removal of the intervention would be impossible (e.g., when skills are taught as part of the intervention), the changing-criterion design achieves the same goal not by withdrawing the intervention, but by changing the criterion for the target metric and examining response to the change. The changing-criterion design is best suited for demonstrating gradual changes over time (Kazdin, 2003) and allows for the evaluation of an intervention that cannot be removed.

Single case methodology in pediatric psychology. The single case design is ideal for addressing clinically relevant questions with a high degree of confidence in the effects of the experimental manipulation and a low number of participants, given the relatively low base rates for many pediatric disorders, and for CF in particular , the single case design is an ideal fit (Linscheid, 2000). Further, the use of the single case design with a small group of heterogeneous individuals serves to strengthen the argument that the change resulted from the intervention, rather than an extraneous variable, and provides evidence in support of a robust treatment effect. Given the low numbers and relative demographic heterogeneity in the population of families served CF clinics, the single case methodology is ideally suited to the examination of an intervention in such a setting.

Summary: Early Nutrition Intervention for Young Children with CF

This review highlights the need to improve nutrition in children with CF. Ensuring adequate nutrition early in life is critical to promoting and preserving health in CF, but children with CF are at a serious nutritional disadvantage as they burn calories at a higher rate than their healthy peers. Despite recommendations from national sources and individual treatment providers to maintain weight at or above the 50th percentile, 23% of children with CF are below the 10th percentile (Stallings et al., 2008). The achievement and maintenance of good nutrition early in life are critical as weight during the toddler and preschool years independently predicts lung function at age six (Konstan et al., 2003; Lai et al., 2009). Researchers have found that one barrier to adequate nutrition is problematic family interactions at mealtime that incentivize and encourage non-eating rather than eating behavior (Stark, 2003b). Research has shown that surgical and education-only nutrition interventions are not as effective at improving long term nutrition and body weight as behavioral interventions (Jelalian et al., 1998; Powers et al., 2003;

Powers, Jones et al., 2005), but efficacious behaviorally based early intervention programs targeted for toddlers and preschool children are not available. Further research is needed to develop and test early interventions to move the field closer toward widespread dissemination of these potentially life-extending interventions.

Statement of Purpose

CF is a serious, life-limiting, genetic disorder that affects respiratory, digestive, reproductive and endocrine systems. Families of children with CF must manage an arduous and complex treatment regimen to maintain health and preserve lung function. One critical aspect of promoting and preserving health in CF is ensuring adequate nutrition early in life, but children with CF are at a serious nutritional disadvantage as they burn calories faster and absorb nutrients more poorly than their healthy peers. The achievement and maintenance of good nutrition early in life are critical as weight during the toddler and preschool years independently predicts lung function at age six. Researchers have found that one barrier to adequate nutrition is problematic family interactions that occur at mealtime. These interactions are likely due to ineffective feeding strategies used by parents who feel enormous pressure to ensure their children eat as much as possible. However these strategies have been demonstrated to incentivize and encourage non-eating rather than eating behavior. Research has shown that surgical and education-only nutrition interventions are not as effective at improving long term nutrition and body weight as behavioral interventions, but many of the current behavioral interventions are aimed at school age children and are not widely available to the CF treatment community.

The goal of this research is to pilot a developmentally sensitive adaptation of an evidence based behavioral parent training intervention to improve nutrition in toddlers and preschool children with CF. The primary goals of this intervention are to demonstrate efficacy by

increasing body weight (as necessary), improving intake as measured by calories and % RDA, and improving non-weight-gain related child feeding problems. The secondary goals of this study are to develop hypotheses about the effectiveness of the behavioral portions of the intervention using observational coding of videotaped family meals to inform future research about the mechanisms of change.

Hypotheses are stratified by the primary and secondary aims of the study. Primary hypotheses are as follows: 1) children with BMI percentiles below the 50th percentile will increase their body weight at post-treatment and 12-week follow-up; and 2) children will meet individualized nutrition goals (i.e., increased caloric intake, developmental appropriateness) at post-treatment and maintain gains at 12-week follow-up. Secondary hypotheses are that parents and children will demonstrate change on variables chosen based on individualized goals, including parent and child behavior and parent reports of eating behavior.

Method

Participants and Recruitment Procedures

This study involved the participation of four children between the ages of 18 and 36 months and their families. Families with children within the specified age range and who met inclusion and exclusion criteria were identified as candidates from among all patients at the CF clinic at a university medical center in the mid-Atlantic region. Eligible families were sent a letter explaining the intervention, the parameters of the study, and that investigators would be available at clinic visits to answer questions. Copies of this letter and fliers advertising the study were on display in the clinic. Following an initial mailing of recruitment materials, the investigator either called eligible families or approached them at a clinic visit to answer questions and determine interest. The study was approved by the local IRB.

Inclusion and exclusion criteria. Inclusion criteria were (1) confirmed diagnosis of CF, (2) between 18 and 60 months at the beginning of the intervention, (3) diagnosis of CF for at least 3 months, and (4) on an unrestricted fat diet.

Exclusion criteria were (1) formal diagnosis of developmental delay (e.g., autism, mental retardation), and (2) diagnosis of another disease or condition known to restrict growth (e.g., history of bowel resection or short bowel section, genetic disorders related to growth).

Participants. Of the 11 families whose children met the eligibility criteria, seven declined to participate, citing time (n=3), distance (n=3), and no current behavioral problems (n=4) as reasons. One parent did not return telephone calls. The intervention was delivered in two groups at two different times to increase recruitment. Families of four children (three boys) with CF participated in the intervention, two in each group. The Hollingshead (1975) two-factor index of social status (education and occupation) was used to classify family socioeconomic status (SES) on a 1-5 scale; families were either 3 (skilled craftsmen, clerical, sales workers) or 4 (minor professional, technical workers). All parent participants were Caucasian and married; all children had pancreatic insufficiency and required pancreatic enzymes for proper digestion. One child in each group had a body composition measure below the 50th percentile at baseline.

Table 1

Participant demographics

	Group 1		Group 2	
	Anna	Ben	Chris	Peter
Age at intake (months)	25	27	30	21
Gender	Female	Male	Male	Male
Participant	Mother, Father	Mother, Father	Mother	Mother
SES	3	4	4	4
BMI Percentile	91.6%	26.0%	2.9%	
WTL Percentile				92.0%

Note. SES = Socioeconomic Status, BMI = Body Mass Index, WTL = Weight to Length.

Measures

Anthropometric measures. Anthropometric measures such as height and weight were collected within two weeks of the beginning of the intervention, at post-treatment and at the three-month follow up. If families did not have an appointment at the CF clinic within that time period, they were asked to obtain these measurements from their regular pediatrician and report them to the principal investigator (PI). These measurements allowed for the calculation of height percentile for age, weight percentiles for age and height, BMI percentiles, percentage of ideal body weight attained, and comparisons to same age peers using standardized population benchmarks available through the Centers for Disease Control (CDC, 2011). For children age two years and younger, weight to length (WTL) percentiles are calculated as an index of body composition; for children older than two years, BMI percentile is used.

Children's ideal body weight (IBW) was calculated by determining the child's "height age," (i.e., the age at which the child's current height would be at the 50th percentile) and then identifying the corresponding 50th percentile weight value for that height. This value was used to calculate recommended daily allowance (RDA; equal to 102 calories per kilogram of IBW).

Diet diaries. Parents were asked to complete 3-day diet diaries each week to document the target child's food and liquid intake. Parents were given digital kitchen scales (EatSmart Decision Pro Digital Kitchen Scale), dry and liquid measuring cups, measuring spoons, and handheld calculators. The first meeting of the intervention focused on accurately measuring and recording children's consumption of various foods (e.g., cereal, home-made recipes), and families practiced using measuring equipment and diet diaries with the assistance of the PI and RD (group #1) or assistant therapist (group #2). Further, PI checked in with families each session to elicit questions about the use of diet diaries and recording equipment. Families were given

written handouts with tips, reminders, and shortcuts and also encouraged to bring nutrition labels for any food about which they had questions.

Diet diaries were checked for completion at the beginning of each session, and missing data were elicited and collected. Parents varied in the level to which they completed the diet diaries. Although the importance of knowing calorie estimates for each meal was emphasized, parents were also told that if they were unable to complete the calorie calculations, they should prioritize capturing as accurately as possible the intake data. Parents were also asked to provide information about the method of preparation (e.g., pureed, fried, baked, etc.).

Calorie content for each food was calculated by weight or volume based on the information provided on the package. In lieu of readily available nutrition facts (i.e., at a restaurant), families were asked to record as much detail about the meal as possible and report on the proportion of the serving consumed. Parents were provided with the Calorie King Calorie, Fat, and Carbohydrate Counter (Borushek, 2010) as well as nutrition information for a number of fast food and family dining establishments in the area and were encouraged to calculate calories consumed to the best of their abilities. Parents were also provided with blank recipe forms (see Appendix A) to facilitate data collection for homemade meals. Using information provided on these forms, the PI calculated calories per serving or by weight or volume and returned this information to parents for later use.

Behavioral assessment. Each family was asked to video record one family dinner per week and was provided with a digital video recorder (Flip Ultra Digital Video Camera, Cisco) and a small tripod for the duration of the active portion of the study for this purpose. Families were asked to ensure that the meal was captured in its entirety (i.e., that the family's meal was captured from beginning to end), that the target child was clearly visible on the screen, and that

the meal coincided with one of the diet diary days so that complete nutrition data would be available for the recorded meal. If parents recorded a meal not captured on a diet diary day, parents were asked to provide intake data for that meal.

Videotaped meals were coded using the Dyadic Interaction Nomenclature for Eating (DINE; Stark et al., 2000). The DINE measures three categories of behavior: parent, child, and eating. Parent behaviors included direct (alpha) and indirect (beta) commands, coaxes, reinforcements, and talking. Child behavior consisted of refusals or complaints about food, requests for food, crying, talking, playing, and moving away from table. The majority of parent and child behavior are coded on an occurrence/nonoccurrence basis in 10-second intervals, with the exception of direct and indirect commands, which are coded according to frequency within the 10-s interval. Eating behavior consists of bites and sips and is coded as number of each per 10-s interval. Families recorded one dinner meal each week during the baseline and intervention phases and another during the follow-up week. Families were asked to videotape meals that are part of the three-day diet diary or else record the food eaten at the videotaped meal to allow for calculation of calories per bite and sip.

Behavioral Pediatrics Feeding Assessment Scale (BPFAS; Crist & Napier-Phillips, 2001; Crist et al., 1994) is a 35-item questionnaire that examined parent and child behavior associated with poor nutritional intake and was used to assess parent report of children's mealtime behavior. The first 25 items assess child behaviors, and the last ten focus on parent behaviors. Parents report on the frequency of each item using a Likert scale (1 = never to 5 = always) and also report on whether the particular behavior is a problem for the family using a yes/no format. The BPFAS yields four scores: Child Behavior - Frequency, Child Behavior - Problem, Parent Feelings/Strategies - Frequency, and Parent Feelings/Strategies - Problem. The

frequency scores reflect how often problematic behaviors occur, and the problem scores indicate the number of behaviors parents consider to be a problem. Higher scores are indicative of maladaptive feeding behaviors and ineffective parental strategies for managing mealtime behavior. Child items include “gets up from table during a meal” and “tries to negotiate what he/she will eat and what he/she will not eat.” Parent items include “I coax my child to get him/her to take a bite” and “if my child does not like what is being served, I make something else.” The BPFAS was developed to measure mealtime behaviors in young children (age 9 months to 7 years, Crist & Napier-Phillips, 2001) and has demonstrated adequate internal consistency for the measure as a whole and the behavior domains ($\alpha = .76-.88$), strong temporal stability reliability in samples not participating in an intervention ($r = .82-.85$), and good construct validity (Powers et al., 2002).

Intervention

The combined behavior and nutrition group intervention targeted one meal and one child behavior management skill each week to foster gradual changes in eating and behavior. This intervention was based on Stark’s 8-week behavior and nutrition parent intervention developed for school age children (Be In CHARGE; 2003a). Developmentally sensitive adaptations were guided by Satter’s (2000) work feeding toddlers and preschool children. Treatment occurred over an 8-week period and included a baseline study visit (week 1) and 6 intervention visits held in weeks 3 to 8. Each session included both nutritional counseling and behavioral treatment components. A group approach was used, and detailed session content is available in Table 2. Overall, the treatment curriculum focused on 3 areas: (1) increasing calorie and fat intake, with a goal of meeting the 120% to 200% RDA for energy, (2) teaching effective parent management skills to address common behavioral challenges of toddlers and preschoolers at mealtime (i.e.,

praising, ignoring, and setting limits), and (3) ensuring appropriate dosage and timing of pancreatic enzyme replacements. Parents were asked to use the behavioral parenting skills regularly at home with all meals and snacks. Using the data from each child’s baseline diet record, individualized goals monitored by a registered dietician were set for energy intake or dietary change.

Table 2

Schedule of Session Content

Week	Session	Nutrition-Focused Topic/Meal Targeted for Change	Behavioral Child Management Technique
1	1	Baseline Teaching: How to keep a food record	Baseline Teaching: How to operate camera to record meals
2			
3	2	How to use snack to meet your child’s energy needs	Differential attention (compliments, praise, and ignoring)
4	3	Breakfast: The most important meal of the day	Contingency management and meal duration
5	4	Relax, review, and problem-solve	Refining use of differential attention and limit setting
6	5	Lunch: Consistency across situations	Limit setting: How to do it and do it well
7	6	Dinner: Making it a family effort	Bringing all the skills together
8	7	Review: Putting it all together and sick days	Review and problem solve: Making behavioral techniques work for you

Timing of Assessments

Questionnaire and anthropometric data at baseline (week 1), post treatment (week 8), and follow-up (week 20) to allow for comparison from pre- to post-treatment and to measure the retention of treatment gains. Families completed three-day diet diaries and video taped one family meal each of the eight weeks of the intervention and the follow-up week.

Participant reimbursement. Given the high data collection burden placed on families participating in this trial, families were reimbursed with gift cards from local supermarkets for

the return of data on a weekly basis. The regular provision of gift cards for data collection served two purposes: first, it incentivized data collection and compensated for families' time, and second, it provided a relevant example of some of the behavioral principles (e.g., reward and reinforcement) covered in the parent training intervention. Parents were given \$5 for each set of diet diaries and \$5 for each video recorded meal each week during the baseline and active treatment phases. Parents were given \$20 for the completion of all measures and assessments at the follow-up time point.

Video Coding and Reliability

Mealtime behavior. Each recorded meal was viewed at least 3 times by an observer using the DINE. Parent behavior was coded during the first viewing, eating behavior was coded during the second viewing, and child behavior was coded during the third viewing, consistent with the methods described by Stark and colleagues (2000). The operational definitions of the DINE coding are provided in Appendix B. Two observers conducted all of the mealtime coding. The study author was the primary coder; the secondary coder was Ph.D. level psychologist with training in pediatric psychology and cystic fibrosis and extensive research experience with observational coding methodologies. Videos of non-participant families with children in the eligible age range were used for training and reliability purposes. Training consisted of instruction on the coding manual, regular meetings to discuss questions and inconsistencies, and independent coding of the selected criterion videotapes until reliability statistics of $\geq .80$ were achieved for each class of behavior. Several reliability statistics were used: Kappa, ICCs, and percent agreement. Kappa was the most frequently used statistic, employed primarily to evaluate agreement on the occurrence/non-occurrence variables. Percent agreement was employed when the behaviors occurred at such a low frequency that kappa was deemed inappropriate (i.e.,

physical prompt, child request; Nelson & Cicchetti, 1995). For the bite, sip, spit-up, and feed data, in which raters were coding frequency of behaviors within each 10s interval, ICC(2,2) was used to calculate agreement (Shrout & Fleiss, 1979). Following the training phase, the primary rater coded all videos, and the secondary rater coded a subset of videos (25%, $n = 9$) to ensure reliability.

Treatment Fidelity. A treatment adherence checklist was created to measure the primary behavior (e.g., planned ignoring, labeled praise) and nutrition (e.g., specific recommendations for each meal) elements. A copy of this checklist is included in Appendix C. This system seeks to demonstrate fidelity to the treatment manual by providing evidence that no drift took place. Each group intervention session was digitally recorded in its entirety, and these fourteen recordings were renamed prior to coding to ensure the coder would remain blind to the session number. A masters-level graduate student with extensive training in observational coding of therapeutic interventions blind to the treatment and hypotheses completed the adherence coding.

Results

Overview

This study provides a preliminary single-case series test of an adapted manual-based treatment for parents of toddlers and preschool children with CF. Results are presented here as follows: (a) missing data and data coding procedures are discussed and (b) outcomes are reviewed for each participant, with primary outcomes presented first.

Sample. Four families participated in the intervention and completed the study, two families in each of two groups. The first group consisted of Anna and Ben's parents and the second group consisted of Chris' and Peter's mothers. All of the children's and parent's names have been changed to protect their confidentiality. The first group was lead by the PI. The CF

Clinic's registered dietitian (RD) attended the first meeting of the first group to facilitate questions regarding food measurement and diet diary completion. The second group was lead by the PI with assistance from a master's-level clinician; due to time constraints, the RD did not attend the first session of the second group. However, all nutrition recommendations were individually reviewed and approved by the RD prior to group meetings, and throughout the intervention.

Treatment goals and outcome measurement. Goals of the intervention were tailored for each child; two children, Ben and Chris, were both below the 50th percentile for body composition and had goals to (a) increase caloric intake and (b) increase body weight. However, Anna and Peter were above the 80th percentile and had different goals. Anna's goal was to improve the nutritional quality of her food (e.g., replace processed gummy snacks with raisins). Given his diet of primarily spoon-fed pureed food, Peter's goal was to replace pureed food with developmentally-appropriate options (e.g., finger food, varying textures). More information about each child and their individual goals is presented later. Due to the substantial differences in goals across participants, results for each child are presented individually.

Primary and secondary outcome measures were selected based on the child's individual treatment goal(s). Primary measures directly assessed progress toward the primary treatment goal; secondary measures were selected to provide additional information about the child's progress on measures relevant to the primary goals of the intervention as originally designed. For example, anthropometric data are presented for both children who did not have weight gain as a therapeutic goal given the focus on body composition for children with CF.

Table 3

Treatment goals and outcome measures

Participant	Treatment Goal	Outcome Measures	
		Primary	Secondary
Group 1			
Anna	Improve nutrition quality	Diet Evaluation	Parent Behavior Anthropometric
Ben	Gain weight	Anthropometric	Caloric Intake BPFAS
Group 2			
Chris	Gain weight	Anthropometric	Caloric Intake Child Behavior
Peter	Developmentally appropriate food	Diet Evaluation	Caloric Intake Anthropometric

Note. BPFAS = Behavioral Pediatric Feeding Assessment Scale

Missing Data Procedures

Diet Diaries. There are two types of missing data on diet diaries; the first more rare and serious category consisted of missing food or amount data. This includes meals wherein no food was recorded or where no amount was provided. In this situation, there is not enough information to reconstruct the meal with the parent and to reasonably estimate the child's intake. As such, this type of data is considered irretrievable and truly missing. For this study, one participant accounted for all of this type of missing data: Chris was missing data from two days in week four, approximately 7% of his daily meal data.

In other cases, the diet diaries contained inadequate information to estimate desired variables without additional calculations. For example, a parent may have forgotten to calculate calories consumed for an item, or they may have provided information that required the investigator to seek out additional information prior to accurately determining the amount of calories consumed. For this study, this type of missing data was much more common, occurring

for approximately 12% of items reported by parents. In the overwhelming majority of these cases (98%), no additional work was required to obtain information to calculate calories consumed. However, a small proportion of data (2%) required additional information to estimate calories consumed by the target.

To estimate these missing data points, the investigator was required to obtain additional information about the overall caloric value of the entire recipe (i.e., food prepared from a mix) or item (i.e., grocery store bakery item) listed on the diet diaries. This required either preparing the item as described by a parent (e.g., a boxed pasta dinner by the instructions provided) to obtain a total weight of all servings as prepared, or purchasing an identical item (e.g., supermarket carrot cake) to obtain a total weight. This additional information allowed the investigator to estimate the caloric value of a food item based on volume, weight, or proportion of the whole.

Anthropometric Data. Anthropometric data were collected (a) within two weeks of the first week of baseline, (b) during the last week of treatment, and (c) during the follow-up week. Each participant had three potential anthropometric data points. Two participants had missing anthropometric data, in both cases due to missed appointments. For these missing data points, anthropometric assessments were collected as soon as feasible and estimates were calculated for the actual target dates using participants' growth and gain rates. These imputed data points were calculated only for illustrative purposes and are clearly identified on figures in which they are used.

Coding

Mealtime Behavior. Videos were coded for behaviors beginning in the interval immediately following the child's ingesting pancreatic enzymes. Kappa coefficients were calculated on exact agreement on the occurrence of behaviors within each 10-second interval

with the exception of bites, sips, and feeds, for which intraclass correlation coefficients (ICCs) using a two-way random model were calculated. Kappa coefficients range from 0 to 1.00, and coefficients of .60 are considered acceptable. The average kappa coefficient was .82 (range, .76 to .92) for parent behaviors, and .83 (range, .75 to .92) for child behaviors. ICCs range from 0 to 1.00, and correlations of .60 are considered acceptable. The average ICC(2,2) for eating behaviors was .91 (range = .89 to .94). All reliability statistics are presented in Table 4.

There were several cases in which kappa could not be calculated, for example, the observed behavior occurred in fewer than 5% of intervals or the occurrence was observed only by one rater. In these cases, percent agreement was calculated (Lagenbacher, Labuouvie & Morgenstern, 1997), the mean agreement was .99 (range = .96 to 1.00). Categories of behavior in which percent agreement was used to calculate agreement varied on a tape-by-tape basis.

Treatment Fidelity. Intervention sessions were coded for presence of nutrition and behavior topics to demonstrate fidelity to the intervention. Fidelity was 100%.ⁱ

Table 4

Agreement statistics for classes of behavior.

Behavior	Kappa		Percent Agreement		ICC	
	n	M (range)	n	M (range)	n	M (range)
Parent						
Command (a)	6	.92 (.80 – 1.00)	2	.99 (.99 – .99)		
Command (b)	6	.87 (.72 – 1.00)	1	.99		
Coax	9	.77 (.62 – .93)	0	-		
Reinforcement	6	.76 (.38 – 1.00)	3	.99 (.98 – 1.00)		
Parent Talk	9	.79 (.65 – .93)	0	-		
Plate Away	2	.90 (.79 – 1.00)	0	-		
Child						
Refuse	9	.83 (.22 – 1.00)	0	-		
Request	0	-	4	1.00 (.99 – 1.00)		
Cry	5	.82 (.60 – .90)	2	.99 (.98 – 1.00)		
Talk	9	.75 (.51 – .92)	0	-		
Play	4	.92 (.84 – 1.00)	1	.96		
Away	3	.92 (.87 – 1.00)	0	-		
Eating						
Bite					8	.94 (.89 – 1.00)
Sip					7	.89 (.81 – 1.00)
Feed					3	.91 (.82 – .96)

Note. ICC = Intraclass Correlation Coefficient. Total n for each behavior may not total 9 due to variability in frequency of overall behavior.

Outcome Results: Participant 1, Group #1: Anna

Anna was a 25-month-old girl at the start of the intervention. Her parents were married and her father attended five out of seven intervention meetings and was regularly involved in her care, particularly at mealtimes. Her mother attended all intervention meetings and was her primary caregiver. She had an older sibling at home. Anna's therapeutic goal was to improve the nutritional quality of her diet; therefore, the primary outcome measure was an evaluation of the elements in her diet targeted by the intervention. The secondary outcome measures included her anthropometrics and her parents' behavior at mealtime.

Primary Outcome Measure: Diet Evaluation

At baseline, Anna's BMI percentile (92nd) put her in the overweight category, and her diet was concerning for its poor nutritional quality. Her caloric intake (mean = 2238) was 140.7% of the RDA (1591) and was not a target of the intervention. Thus, Anna's primary nutritional goal was to gradually replace low nutrient, high-sugar, processed beverages and snacks with more nutritious alternatives. The quantities of low and high nutrient beverages and snacks reported in the 3-day diet diaries are presented in Figure 1. These results demonstrate the quantity of high-sugar beverages and foods and the low quantity of fresh fruit and fruit juice at baseline. The quantity of sugary drinks declined briefly during the first week of the intervention, when the first round of individualized nutrition goals was established. During the same week, consumption of fruit juice increased to its highest point (8 servings) and surpassed sugary drinks. However, in the second week of the intervention, consumption of these foods and drinks returned to near baseline levels. Levels of fresh fruit remained extremely low throughout the intervention, with a peak of three servings per week, despite a focus on replacing sugary snacks with fresh or dried fruit. Dried fruit (i.e., raisins, dried cranberries) was not offered once during the intervention, despite several specific recommendations to replace processed "gummy" snacks with these more high nutrient alternatives. Overall, these data suggest that the intervention was largely ineffective in changing the nutritional quality of Anna's diet.

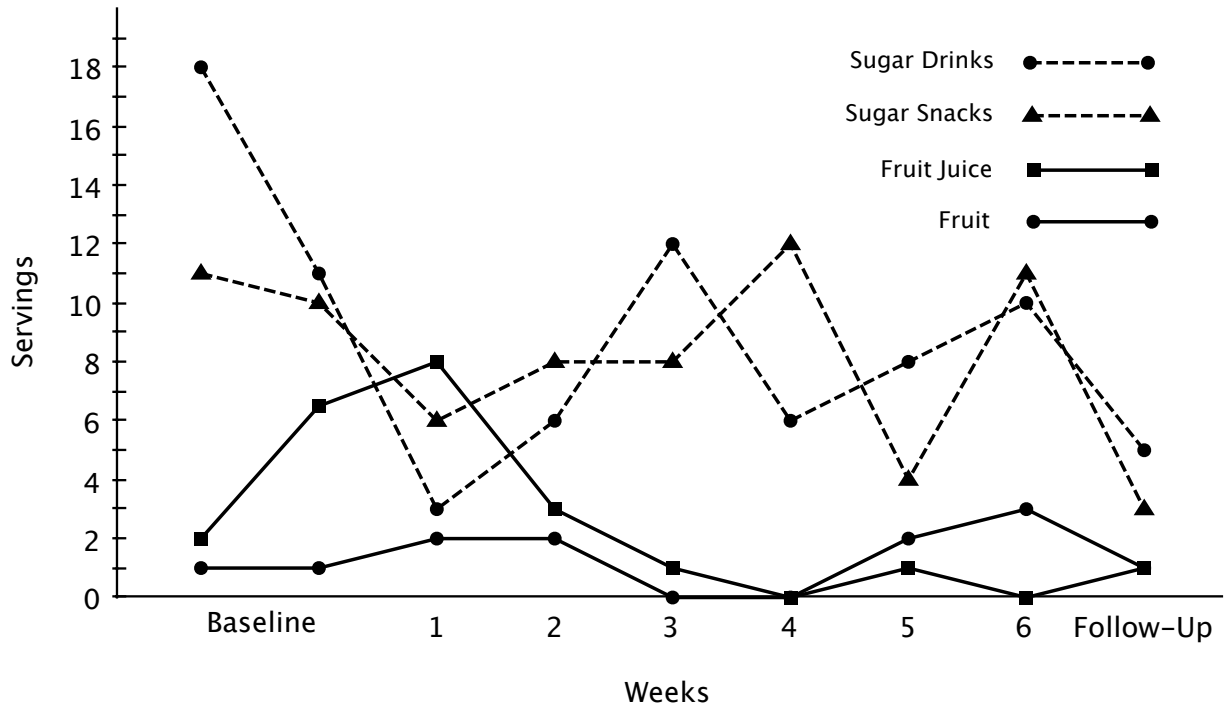


Figure 1. Anna: Quantity of high- and low-nutrient food and beverages offered

Secondary Outcomes: Anthropometrics

Anna's body composition relative to other children her same age is presented in Figure 2. These data demonstrate Anna's history of a consistently high body composition percentile, measured as weight to length ratio prior to 24 months of age and as BMI percentile at two years and older. The star on the graphs represent an imputed data point as Anna's parents were not able to obtain a reliable height and weight measurement within two weeks of the first meeting. As demonstrated, her body composition was consistently in the overweight range, with the exception of the post-intervention assessment, when she was briefly within normal limits. Figures 3 and 4 demonstrate her weight and height on age-normed height and weight curves. These data indicate that whereas her length for age was relatively consistent along the 90th to 95th percentiles since she was 22 months old, her weight showed a steep slope, increasing beyond the 95th percentile.

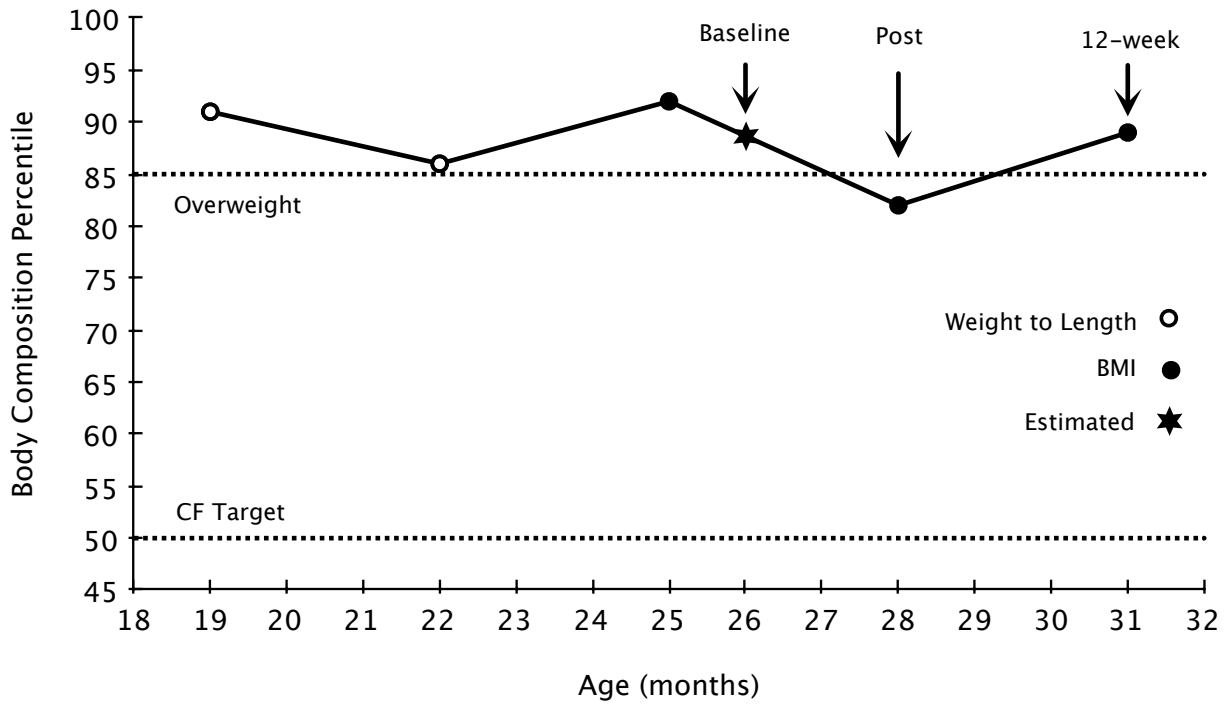


Figure 2. Anna: Body composition percentile

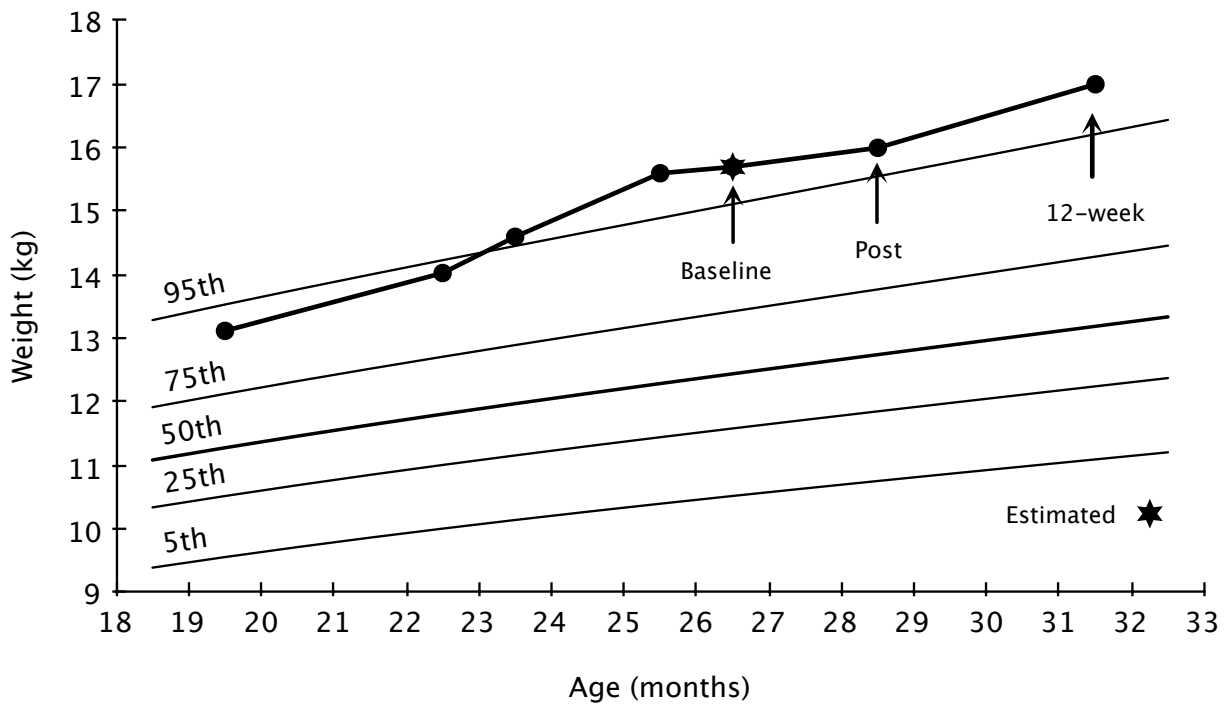


Figure 3. Anna: Weight for age

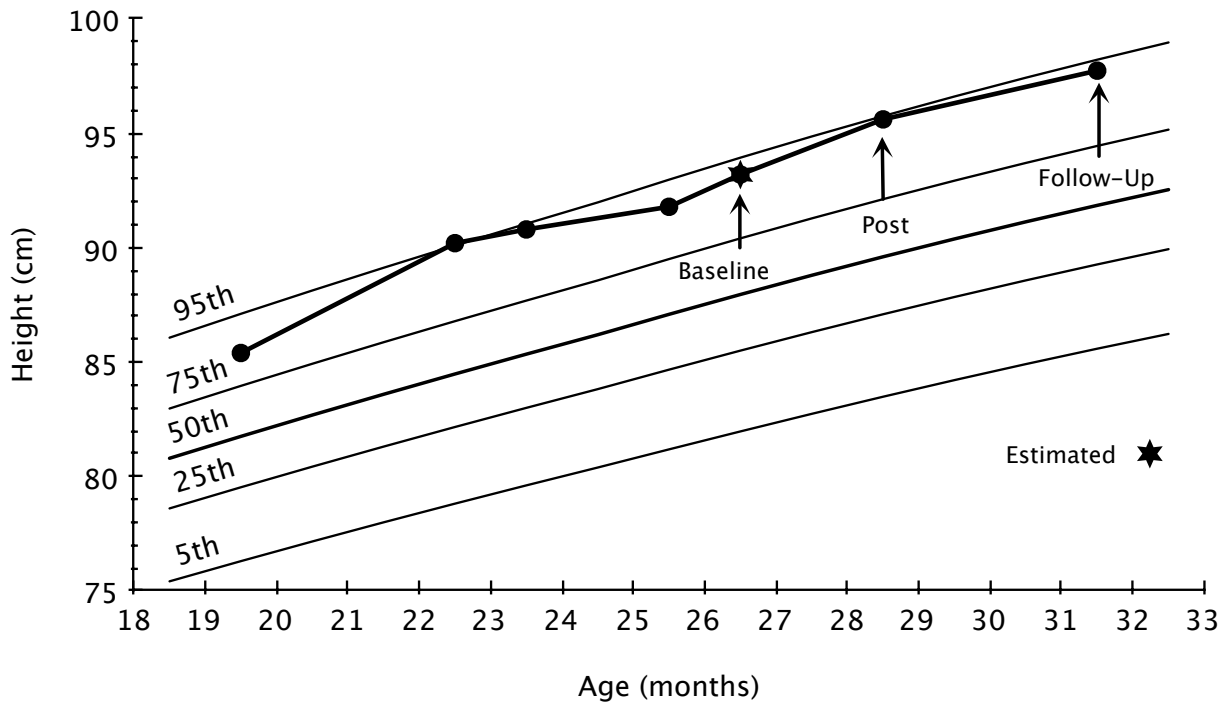


Figure 4. Anna: Length for age.

Secondary Outcomes: Parent Behavior

Despite Anna's already high weight, her parents were highly focused on making sure she ate and spent a great deal of the meal engaged in strategies to increase the amount and speed of her consumption. Parent behaviors gauged by the observational measure include providing reinforcing words or gestures (Reinforce), delivering a clear command to eat (Alpha Command), delivering a command to eat followed immediately by additional words or competing commands (Beta Command), and coercive strategies, including playing physical games, reminding her how good the food tastes, or offering additional or alternate food (Coax). Figure 6 demonstrates the frequencies of these behaviors on an occurrence per minute scale; this scale was chosen to facilitate comparisons across meals of varying lengths. As depicted in Figure 5, Anna's parents demonstrated a high frequency of coaxing behaviors during the mealtime interaction. For

example, they would often pretend or threaten to take food from Anna’s plate to eat themselves or give to her sister, in an effort to eat her food. Alternatively, they sometimes would tell her *not* to eat something in an apparent attempt to coax her to take a bite. In contrast, their rate of reinforcement of her eating behaviors occurred much more infrequently, at a rate of approximately once every three minutes. Anna’s parents also provided direct commands to eat (i.e., Alpha), but they frequently modified such commands by saying something else immediately following the command rather than waiting for Anna to respond (i.e., Beta).

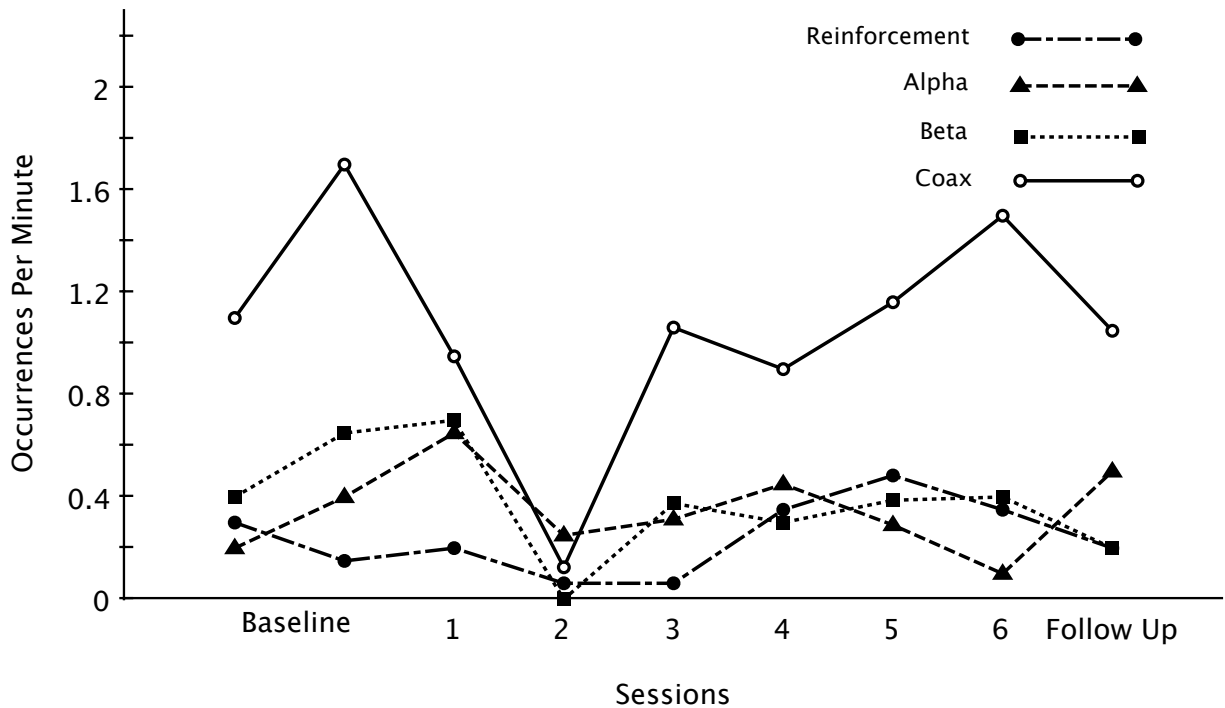


Figure 5. Anna: Parent behavior

Overall, results for Participant 1 offered mixed support for the efficacy of the intervention. The primary outcome, nutritional changes, was largely unchanged. Further, parenting behaviors did not improve during the treatment. However, some positive effects were noted with regard to short-term changes in body composition compared to other children her age.

Outcome Results: Participant 2, Group #1: Ben

Ben was a 27-month old boy at the start of the intervention. His parents were married. His father attended five out of seven intervention meetings and was regularly involved in his care. His mother attended all meetings and was the primary caregiver. Ben has an older sibling from a parent's previous marriage who was in the home half of the time. His primary therapeutic goal was to increase his body weight.

Primary Outcome: Anthropometric

At baseline, Ben was in the 26th percentile for his BMI, well below the 50th percentile recommended by the CFF. Thus, Ben's primary nutritional goal was to increase caloric intake in an effort to increase his weight so his body composition would be above the 50th percentile. Figure 6 demonstrates Ben's body composition across time and his relatively stable history of body composition between the 20th and 30th percentiles. These data also reveal a noticeable increase following the baseline assessment, with a steeper gain in BMI percentile occurring between the post and follow-up assessment, when he achieved the 51st percentile for BMI. Overall, these data suggest the intervention was successful in improving Ben's body composition.

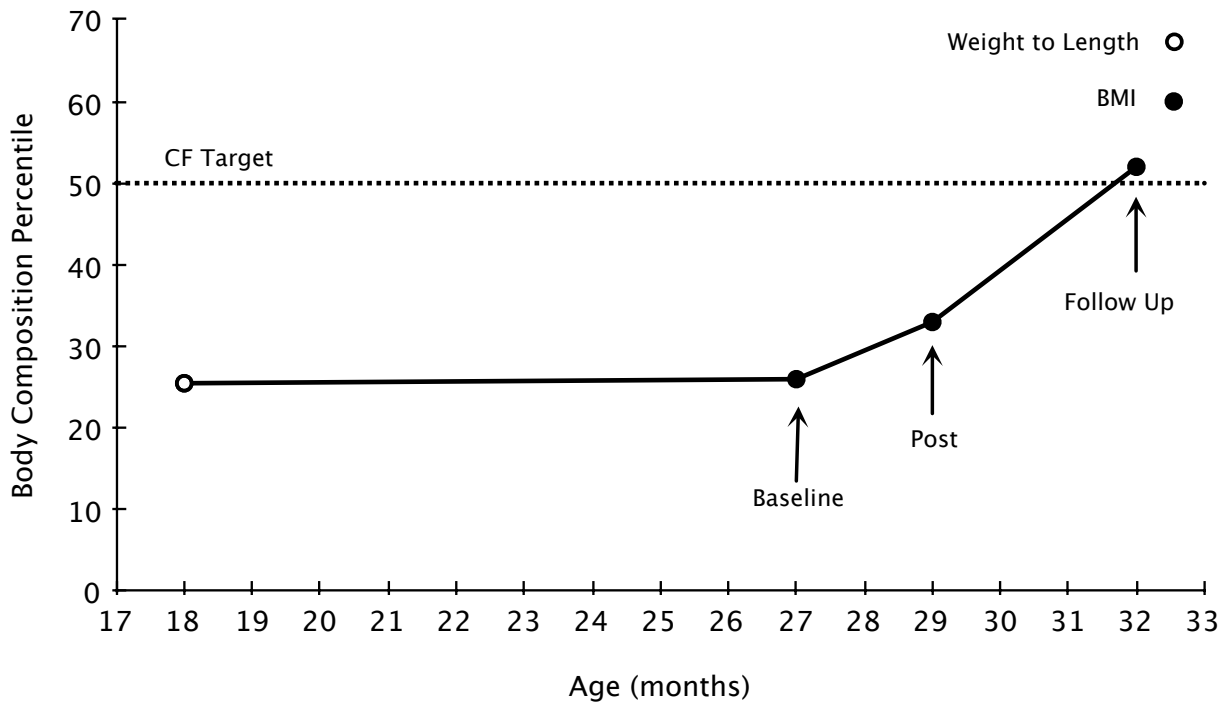


Figure 6. Ben: Body composition percentile

Figures 7 and 8 demonstrate that whereas Ben's relative length for age declined during the intervention, the trajectory his weight for age either remained stable or increased slightly. These data suggest that whereas Ben was experiencing a decline in his rate of growth, he continued to gain weight during the intervention. These data in combination with the increase in Ben's BMI percentile at the onset of the intervention provide evidence of a strong causal link between the intervention and Ben's weight gain.

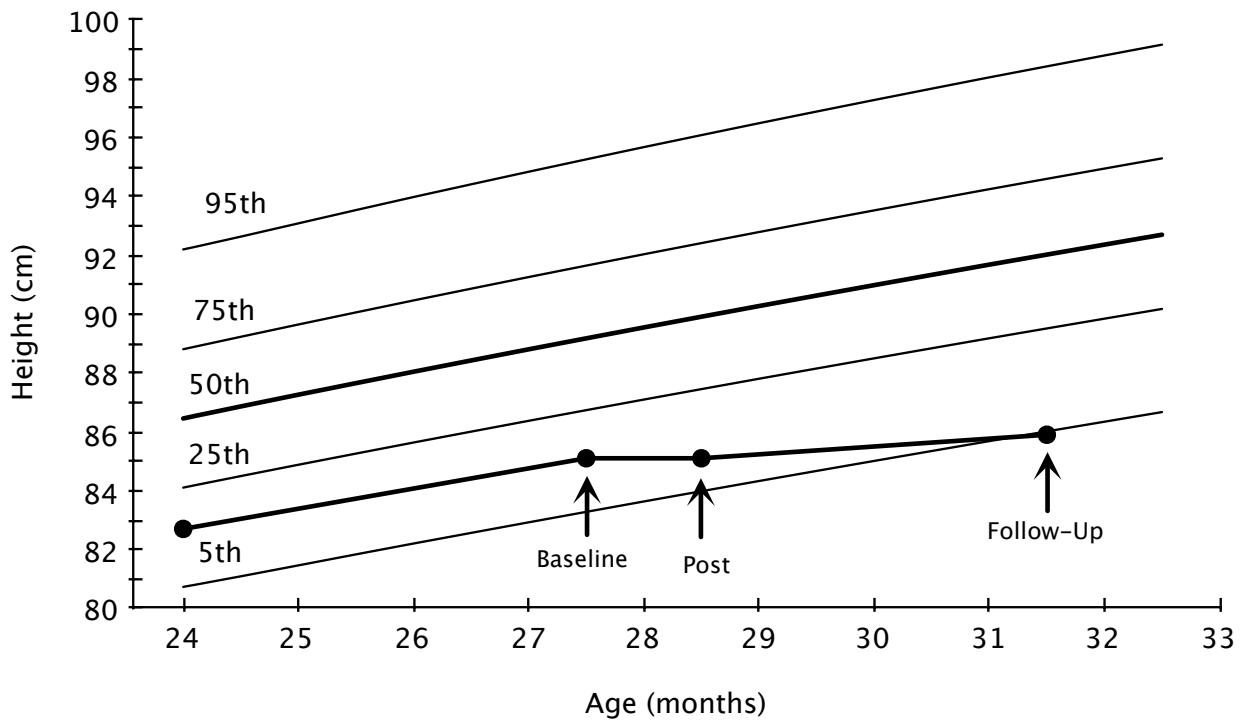


Figure 7. Ben: Length for age.

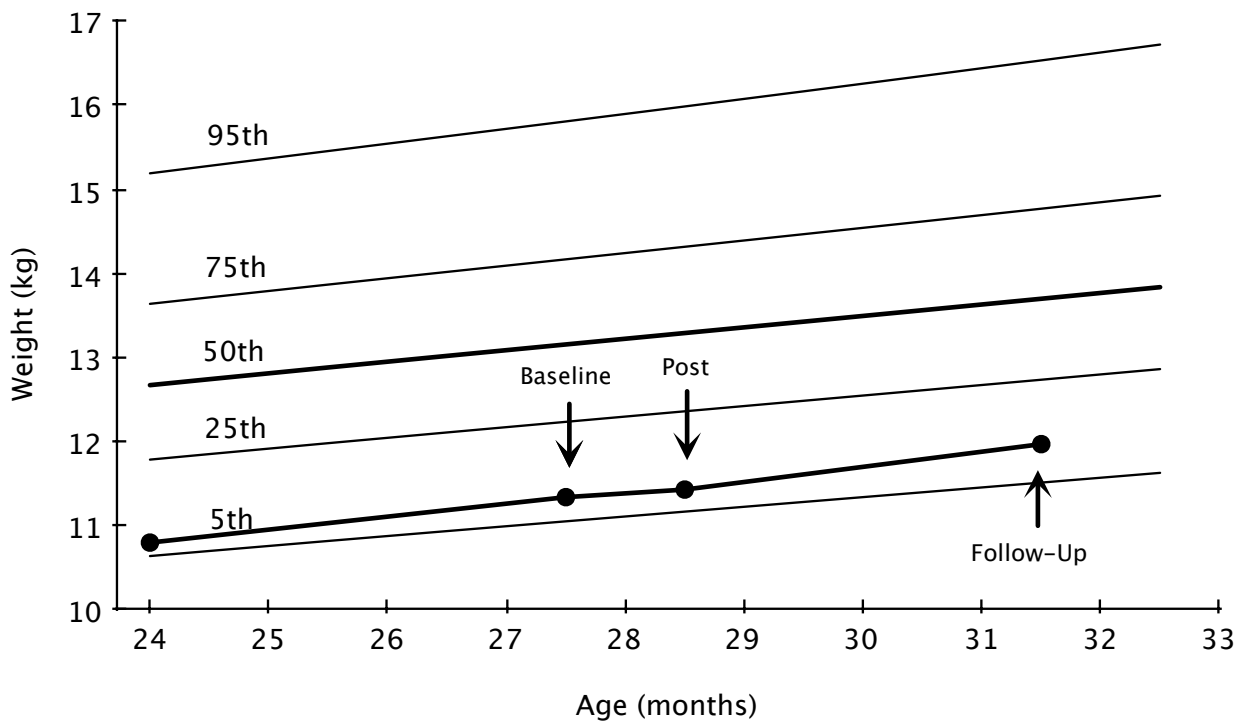


Figure 8. Ben: Weight for age.

Secondary Outcomes: Caloric Intake

Evaluation of Ben's caloric intake includes several variables: (a) total daily caloric intake; (b) daily caloric intake, snacks only; (c) caloric intake daily, by snack; (d) number of snacks offered, daily, (e) daily caloric intake, breakfast only; (f) daily caloric intake, lunch only; and (g) daily caloric intake, dinner only.

Ben's total caloric intake during the two baseline weeks ranged from 750 to 1303 (mean = 1156). This is 92.9% of his RDA, which was 1244 calories/day. Ben's total intake goal was established at 1600 in consultation with the RD, with incremental increases across all four meal categories. This target was established as it falls within the 120-150% RDA range (1495-1865). Table 5 displays Ben's intake per meal across both baseline weeks and the target intake for each meal as well as his total intake.

Table 5

Ben: Baseline and target caloric intake

Meal	Baseline				Target
	Week 1		Week 2		
	<i>M</i>	Range	<i>M</i>	Range	
Total	1122	750 – 1314	1189	1177 – 1209	1600
Snacks	381	0 – 624	414	305 – 480	500
Breakfast	120	90 – 142	118	144 – 210	300
Lunch	310	231 – 440	296	232 – 419	400
Dinner	311	220 – 393	290	243 – 375	400

Figure 9 displays Ben's total daily intake across the intervention. The dark banded area indicates the ideal goal range for his daily intake, between 120% and 150% of his RDA. The short horizontal lines display the average intake across the three-day diet diaries for that week. These data demonstrate that during the two weeks of baseline, Ben's average intake was below his RDA. His overall average intake remains relatively stable for the first four weeks of the intervention. In the fifth week, his average weekly intake exceeds the RDA, and in the final

week, his average intake just falls short of the 120% intake goal. At the follow-up assessment, his average weekly intake is well within the established range. These data suggest that Ben's caloric intake increased as a function of the intervention.

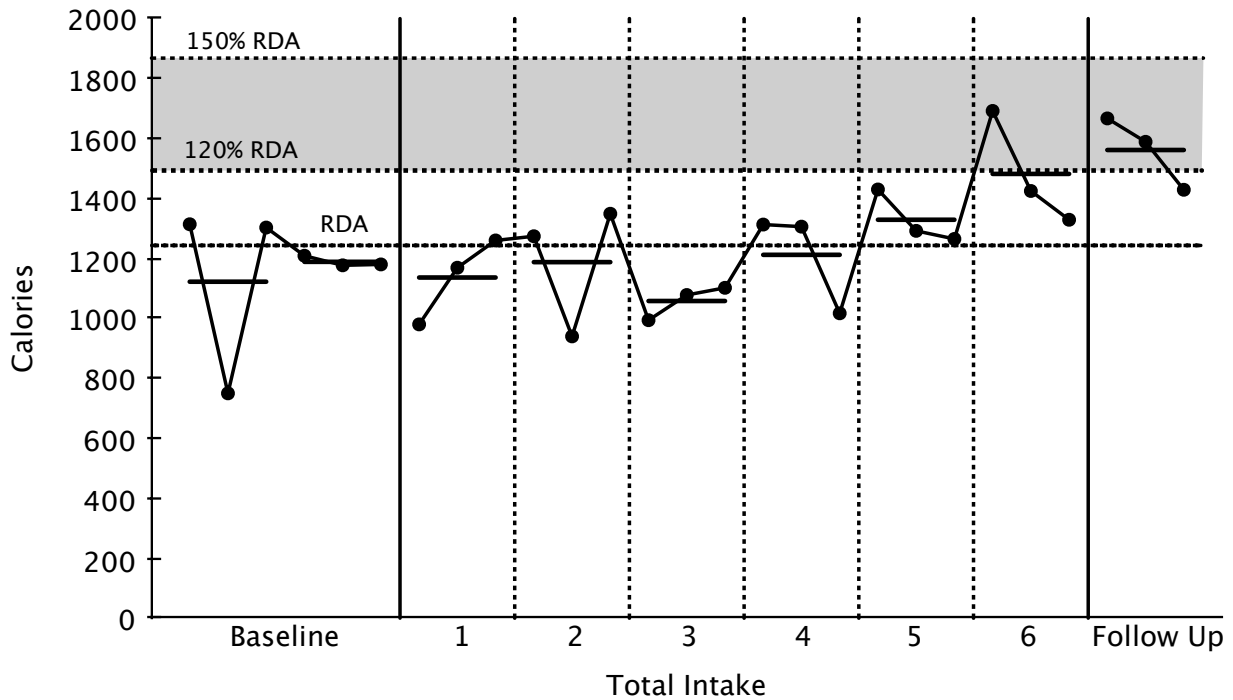


Figure 9. Ben: Total caloric intake across all phases of the intervention. Shaded area indicates CFF target range between 120% and 150% RDA. Short solid horizontal lines indicate weekly means.

Figure 10 displays Ben's total intake at snacks across the intervention. The shaded area indicates the weeks during which Ben's parents were instructed to work toward his snack intake goal of 500 calories per day, indicated by the horizontal dashed line. Snacks were the first meals targeted by the intervention, and Ben was able to meet and exceed the target every week of the intervention. Figure 11 displays the average intake per snack. As snacks differed from the other meals in that they were offered several times per day, understanding Ben's increase in intake requires both an understanding of how calorically loaded each snack was, as well as how frequently snacks were offered (Figure 12). Overall, these data indicate that Ben's increase in

snack calories was due largely to the increased frequency of snacks offered rather than an increase in calories consumed at each snack.

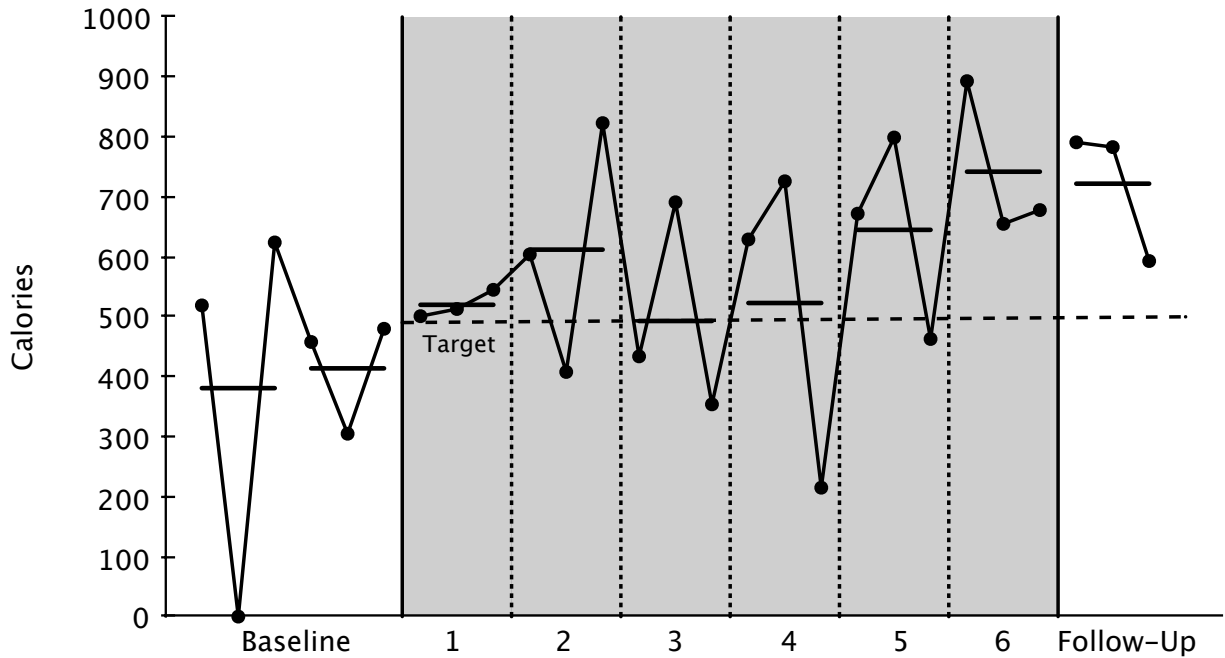


Figure 10. Ben: Total snack calories. Shaded areas indicate times during which snack was a focus of change in the intervention. Solid horizontal lines each week indicate weekly averages. Dashed horizontal lines indicate the target intake level.

The data presented in Figures 11 and 12 demonstrate that the caloric load of the individual snacks offered to Ben did not differ from baseline over the course of the intervention. However, the frequency of snack offerings did demonstrate a visually significant increase.

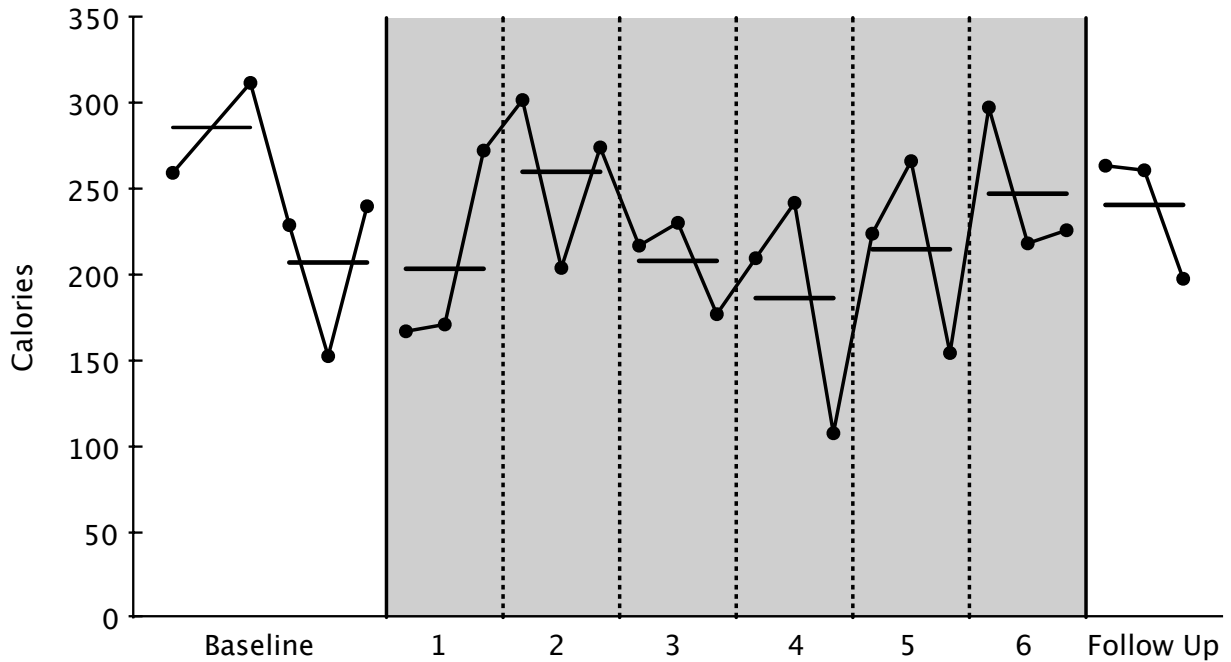


Figure 11. Ben: Average caloric intake of each snack. Shaded areas connote the time during which increasing intake was a focus of the intervention. Short solid horizontal lines indicate weekly means.

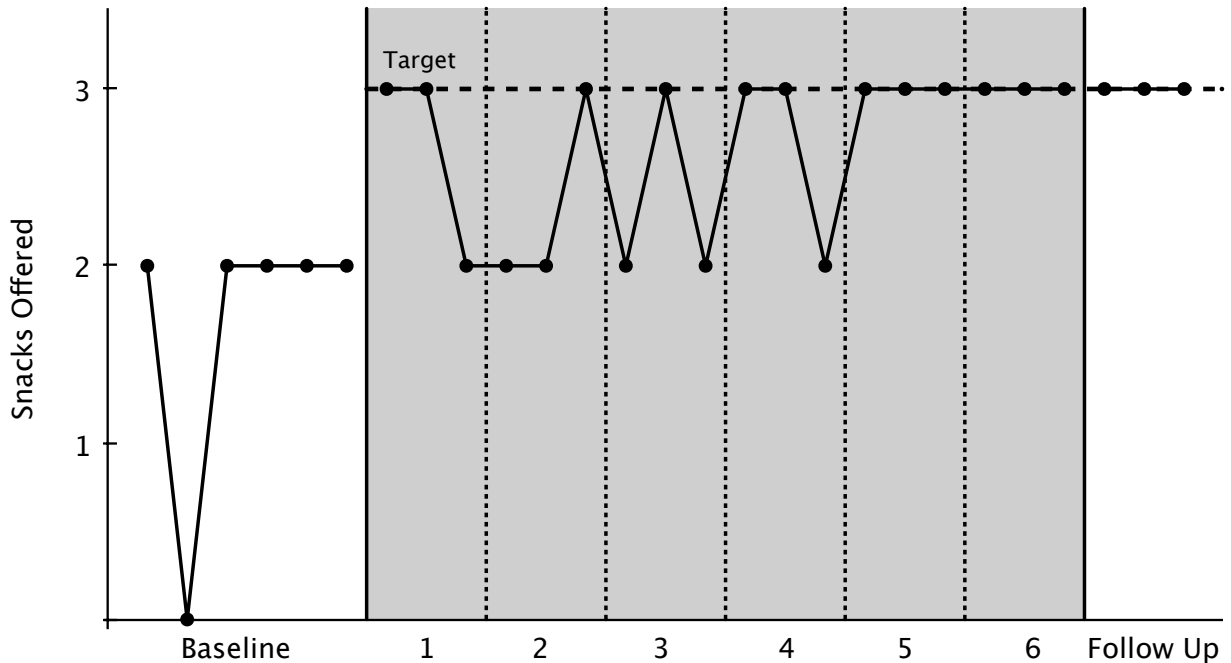


Figure 12. Ben: Frequency of snack offerings. Shaded areas connote the time during which increasing intake was a focus of the intervention. Dashed horizontal line indicates the target snack frequency.

Unlike Ben’s intake at snack, his intake at breakfast did not demonstrate a visually significant increase. The results presented in Figure 13 demonstrate that Ben’s average caloric intake at breakfast remained relatively level throughout the intervention and into follow-up.

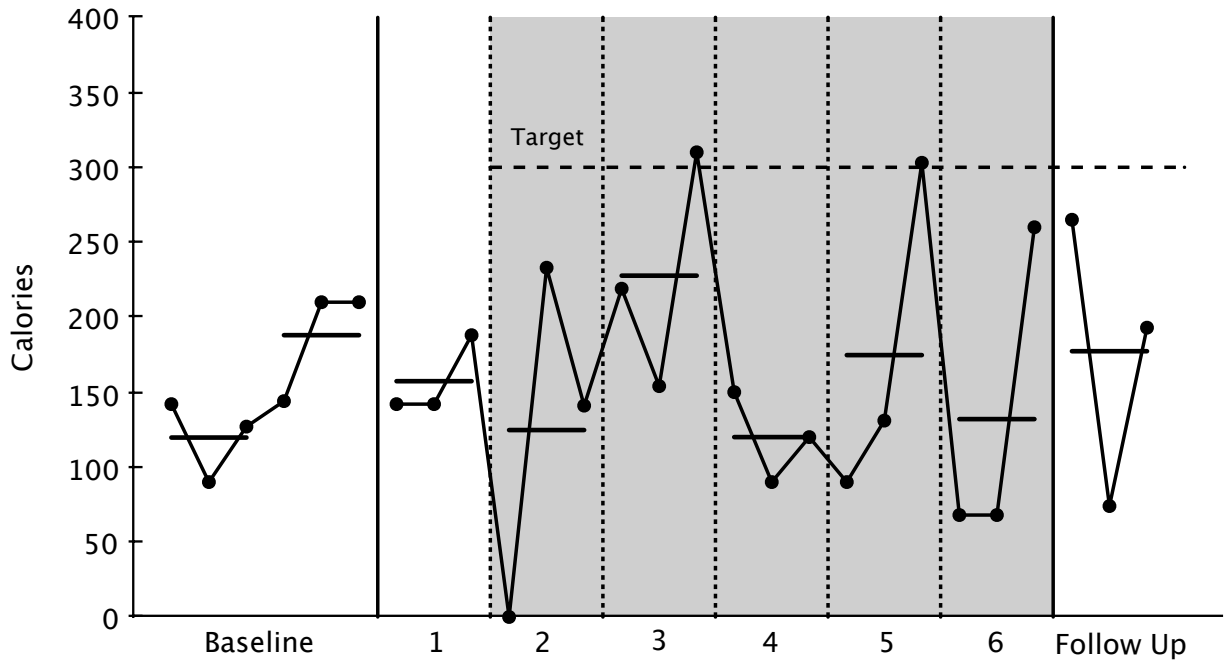


Figure 13. Ben: Caloric intake at breakfast. The shaded areas connote the time during which Ben was meant to consume the target number of calories (dashed line). Short solid horizontal lines indicate weekly means.

Figure 14 demonstrates Ben’s intake at lunch across the phases of the intervention. These data indicate his lowest average lunch intake occurred in week 3, and began to increase in the fourth week, concurrent with the intervention focus on lunch. Ben did not achieve the target for mean caloric intake. He achieved the highest one-day total during the follow up week during which he consumed over 450 calories at lunch. Overall, these data indicate that Ben’s intake at lunch increased as a function of the intervention.

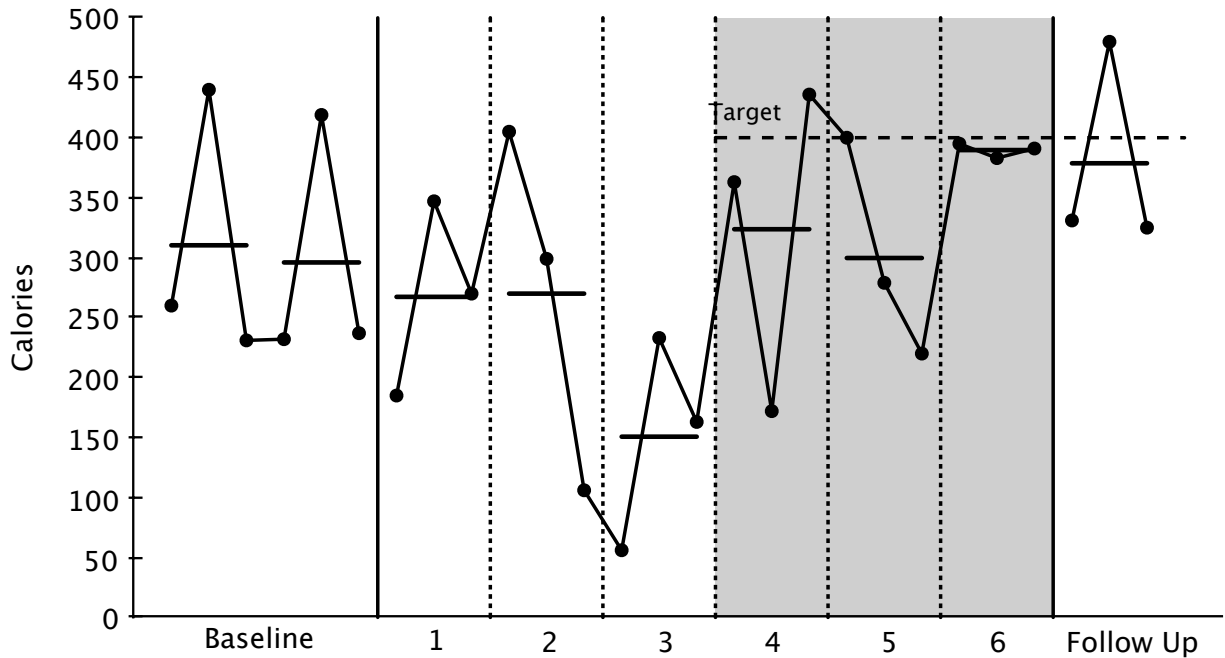


Figure 14. Ben: Caloric intake at lunch. The shaded areas connote the time during which Ben was meant to consume the target number of calories (dashed line). Short solid horizontal lines indicate weekly means.

Ben's intake at dinner is presented in Figure 15. These data indicate that Ben's highest levels of consumption occurred in the baseline phases. Ben's intake at dinner declined during the first week of the intervention, and it remained below baseline levels throughout the intervention. Also of note, Ben had three days during the six intervention weeks during which he did not eat anything for dinner, and he had one dinner during which he ate less than 100 calories. Whereas a visual analysis does suggest a slight upward trend in average intake from week 1 to post and follow-up, Ben's overall intake at dinner remained below baseline levels.

Overall, the individual meal data suggest that whereas Ben's overall intake increased as a result of the study intervention, these changes were not spread evenly across all three meals and snacks.

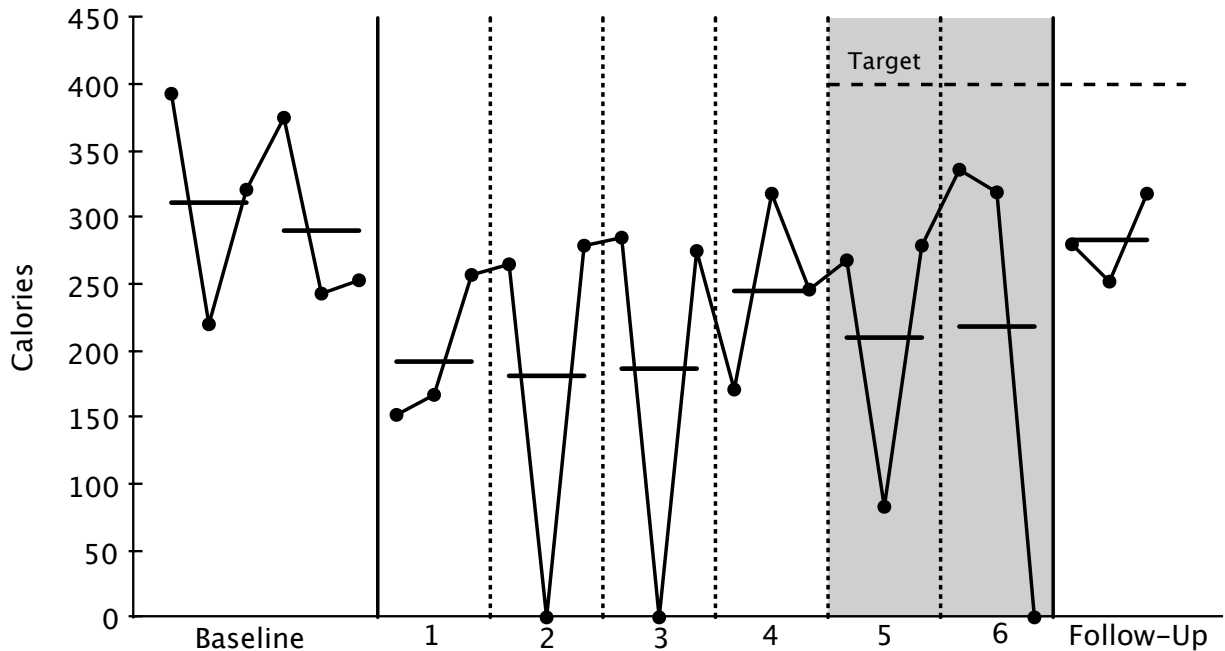


Figure 15. Ben: Caloric intake at dinner. The shaded areas connote the time during which Ben was meant to consume the target number of calories (dashed line). Short solid horizontal lines indicate weekly means.

Secondary Outcome: Parent Report of Mealtime Behavior Problems

Ben’s mother completed the BPFAS at baseline, post, and follow-up, and his father completed the measure at baseline and follow-up. The frequency scores are presented in Figure 16; these scores were consistently below the clinical cutoff and demonstrate that Ben’s mealtime behavior was not clinically significant. The problem scores are presented in Figure 17 and demonstrate Ben’s mother’s perception of Ben’s behavior at both baseline and post-treatment that Ben’s behavior was clinically significant. However, her report of his behaviors at the follow-up decreased below the clinical cutoff. These results indicate that throughout the study period, Ben’s behaviors were within normal limits; however, his mother perceived these problems as highly problematic.

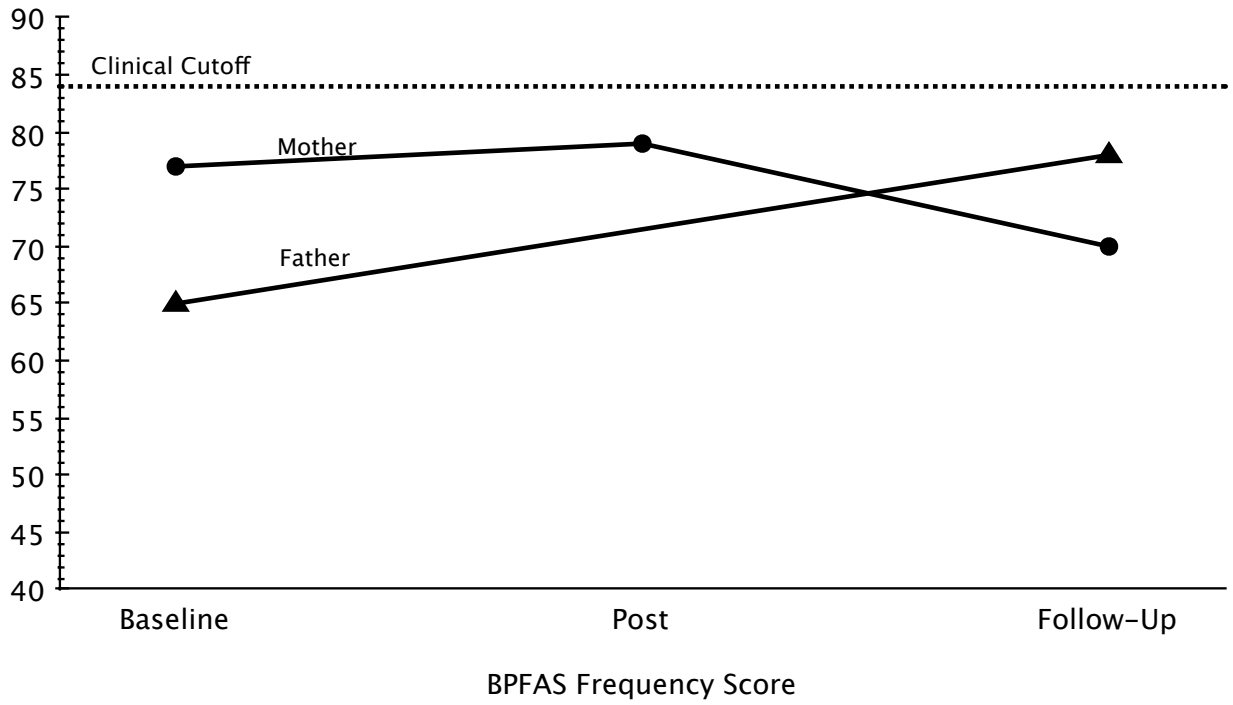


Figure 16. Ben: BPFAS Frequency scores by mother's and father's report.

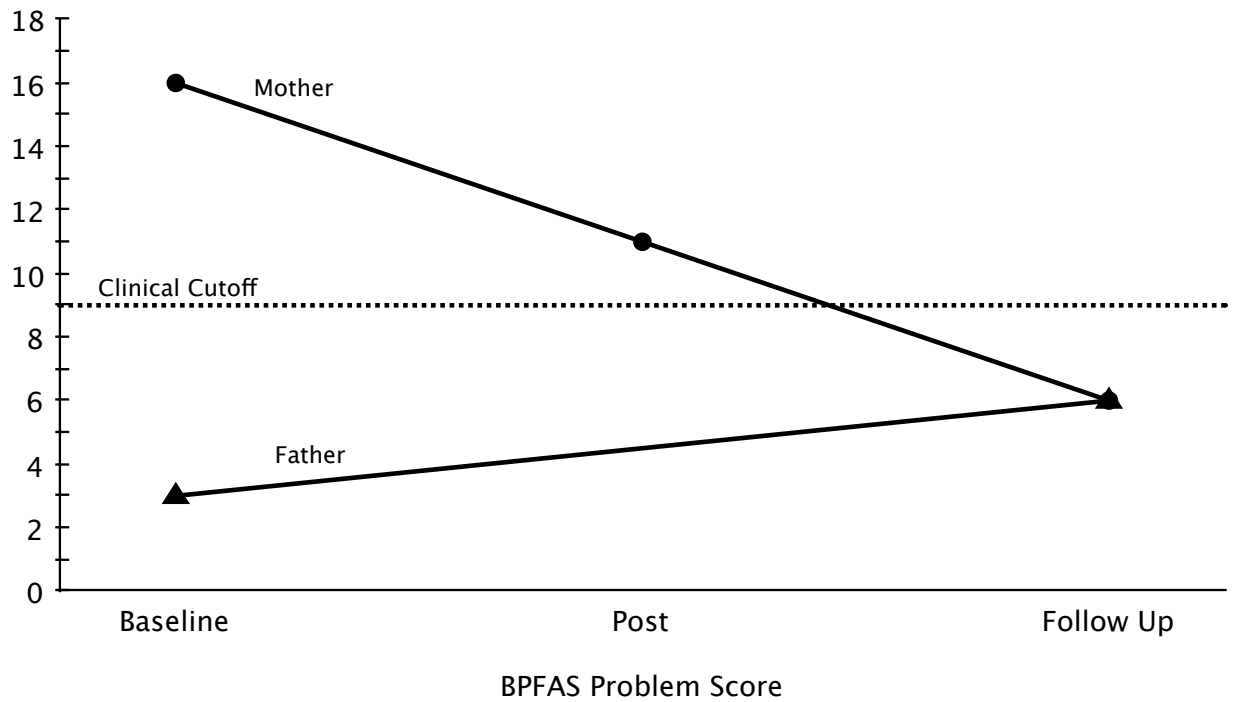


Figure 17. Ben: BPFAS Problem scores by mother's and father's report.

Overall, results for Participant 2 offered strong support for the efficacy of the intervention. The primary outcome, Ben's anthropometric data, demonstrated clinically significant improvement consistent with the onset of the intervention. Further, these improvements continued beyond the termination of the intervention, suggesting retention of treatment gains. Further, Ben's overall caloric intake increased in conjunction with the onset of the intervention, and the follow-up data suggests his increase in intake continued beyond termination. Finally, positive effects were noted in regard to his mother's perception of problems at mealtime, with patterns of improvement noted from baseline to post-treatment continued through the follow-up.

Outcome Results: Participant 3, Group #2: Chris

Chris was a 31-month-old boy at the start of the intervention. His parents were married; however, his mother attended all meetings alone and was the primary caregiver. Chris has a younger sibling with CF and two older siblings from a parent's previous marriage who were intermittently in the home. His therapeutic goal was to increase his body weight.

Primary Outcome: Anthropometric

Chris's weight to length and BMI percentiles across the one year prior to the baseline assessment and continuing to three months beyond the follow-up are presented in Figure 18. Chris's length and weight for age are presented in Figures 19 and 20, respectively. Prior to the intervention, his body composition ranged from the 10th to the 30th percentiles, and at baseline, he was below the 3rd percentile. Three months following the baseline assessment, he reached the 18th percentile, and at seven months after the baseline, he reached the 61st percentile.

Anthropometric data were not available for Chris during the specified time points, so these data

points were estimated for illustrative purposes. Chris’s growth trajectory during the study indicates that the intervention was successful in improving Chris’s body composition percentile.

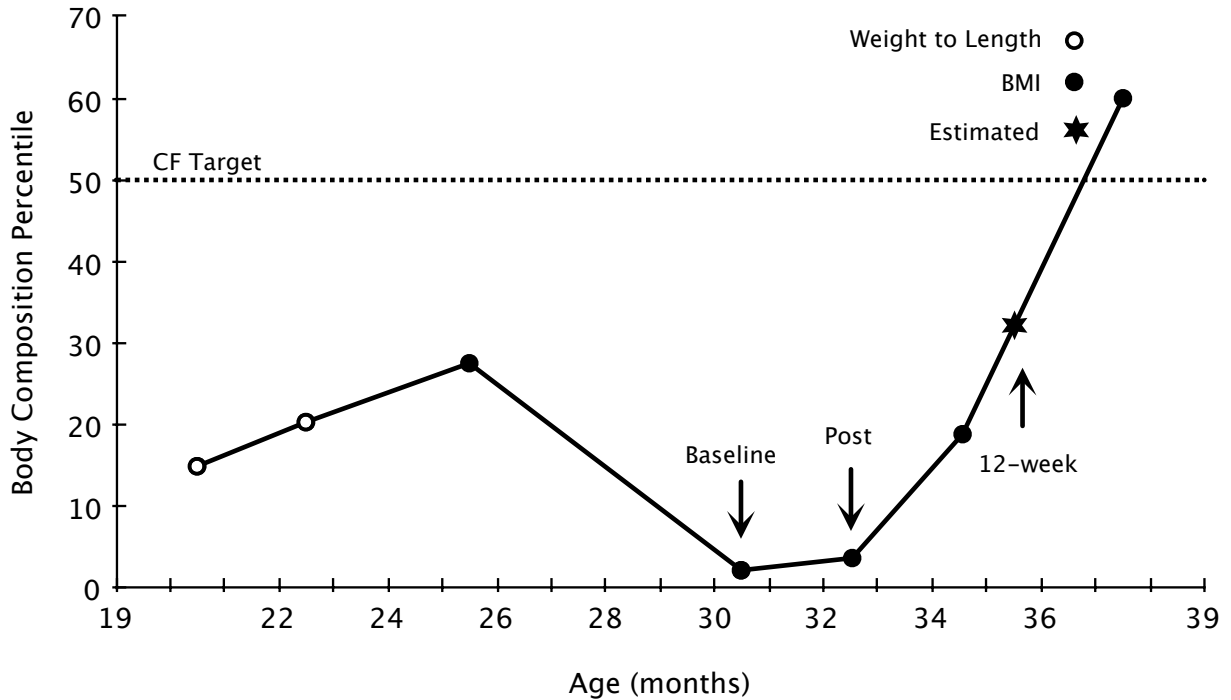


Figure 18. Chris: Body composition percentile.

Figures 19 and 20 demonstrate that whereas Chris’s length for age remained relatively stable between the fifth and 25th percentiles, the trajectory his weight for age reversed at the baseline and began a steady increase. The timing of this change in the slope of Chris’s weight curve indicates that the intervention was causally related to his weight gain.

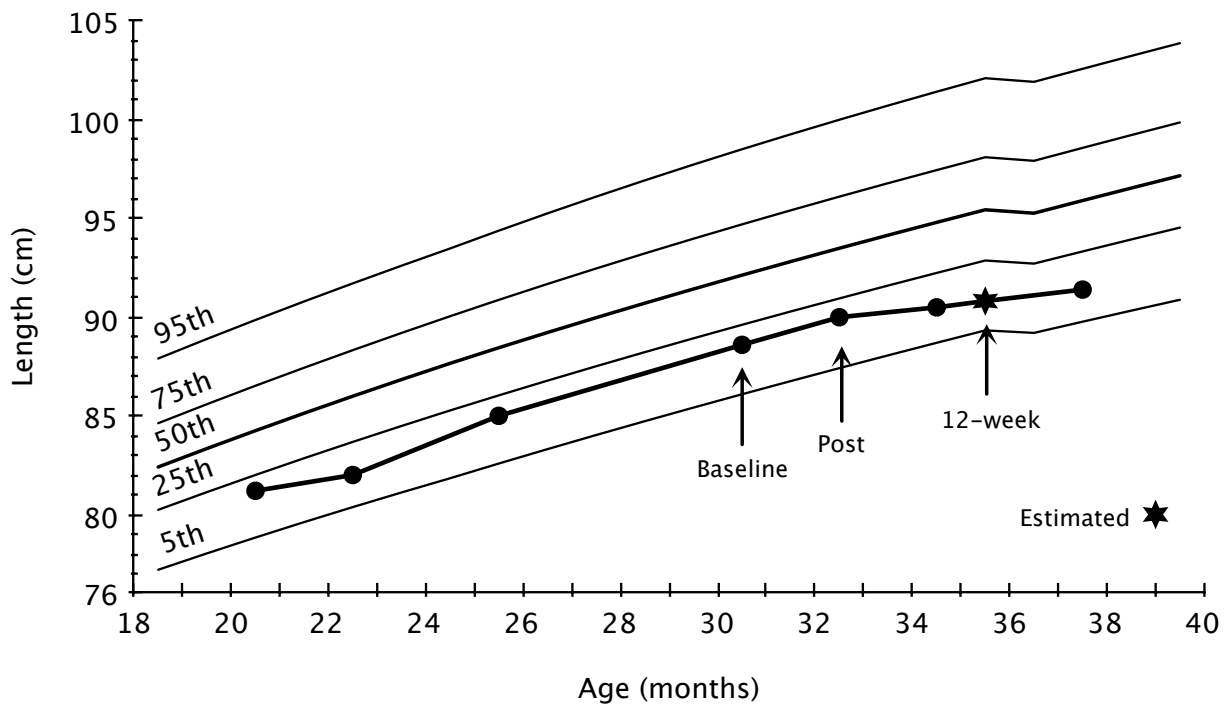


Figure 19. Chris: Length for age.

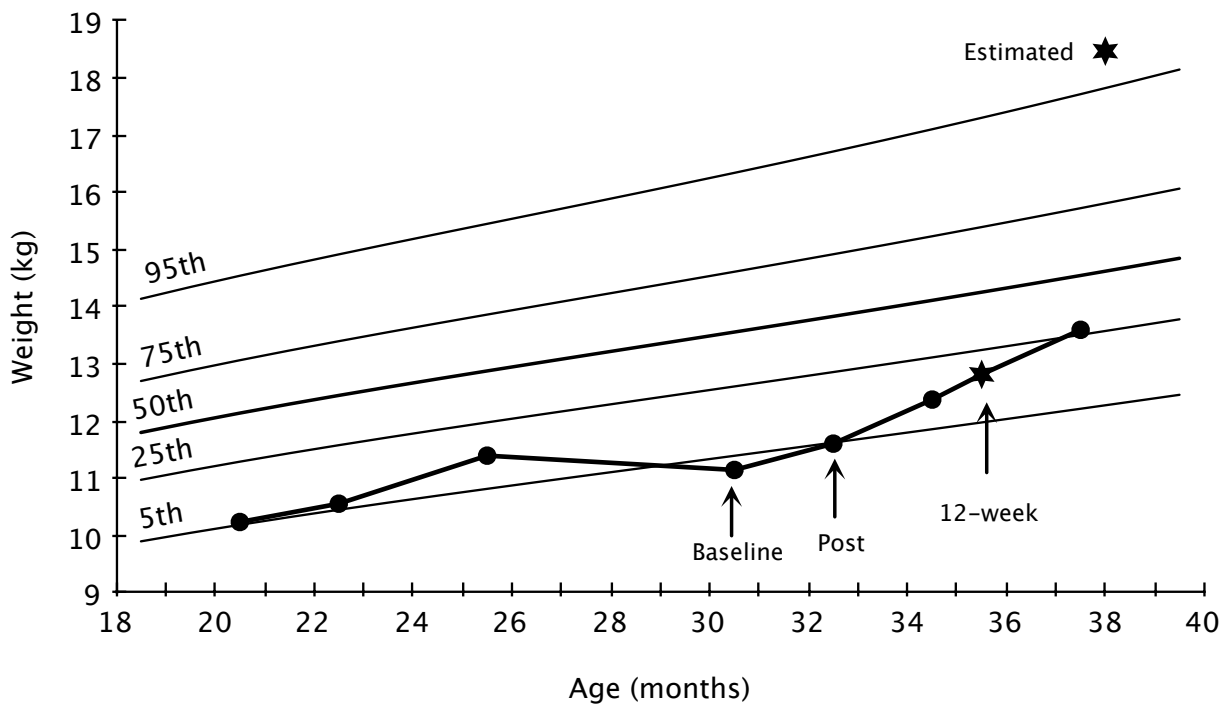


Figure 20. Chris: Weight for age.

Secondary Outcome: Caloric Intake

Evaluation of Chris's caloric intake includes several variables: (a) total daily caloric intake; (b) daily caloric intake, snacks only; (c) daily caloric intake, breakfast only; (d) daily caloric intake, lunch only; and (e) daily caloric intake, dinner only.

Chris's total intake during the two baseline weeks ranged from 1299 to 2093 (mean = 1667). This is 125.7% of his RDA, which was 1326 calories per day. Chris's total intake goal was established at 1900 calories in consultation with the nutritionist and Ben's medical team, with incremental increases across all four meal categories. This target was established as it falls within the 120-150% RDA range (1591 – 1989). Table 6 displays Chris's intake per meal across both baseline weeks and the target intake for each meal as well as his total intake.

Table 6

Chris: Baseline and target caloric intake

Meal	Baseline				Target
	Week 1		Week 2		
	<i>M</i>	Range	<i>M</i>	Range	
Total	1734	1299 – 2093	1599	1439 – 1698	1900
Snacks	474	189 – 634	506	90 – 873	550
Breakfast	354	337 – 384	415	277 – 558	450
Lunch	533	392 – 766	440	323 – 555	450
Dinner	374	311 – 429	237	91 – 349	450

Chris's caloric intake data over the course of the intervention are presented in Figure 21. There were two days of missing data during week two. At baseline, Chris's mean caloric intake was between 120% and 150% of his RDA, a particularly surprising result given his very low body composition. Following the first week of the intervention, his average intake remained above the 150% RDA threshold. However, this increase in intake did not assume the gradual incremental step-like increase predicted.

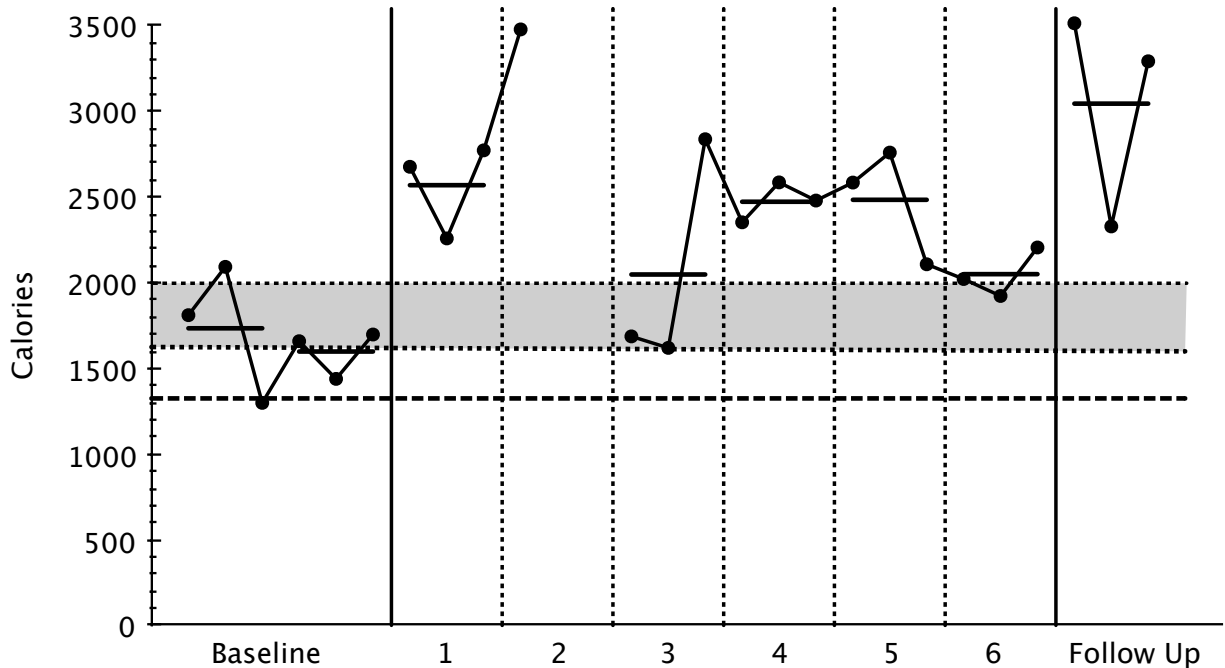


Figure 21. Chris: Total caloric intake across all phases of the intervention. Shaded band connotes 120% to 150% RDA. The dark dashed line indicates the RDA. Short solid horizontal lines indicate weekly means.

Chris's snack intake is displayed in Figure 22. Chris's snack intake dramatically increased following the first week of the intervention. It remained consistently high throughout the intervention and follow-up period. In some weeks, Chris's snack intake was more than double the target, and during the follow up, his average snack intake was three times the target. Overall, these data suggest that the intervention was successful in increasing Chris's caloric intake at snack time.

Chris's caloric intake data at breakfast are displayed in Figure 23. The data indicate that Chris's breakfast intake increased and remained above target levels until the last week of the intervention, dropping just below the target. Chris's breakfast intake was below target levels at follow-up. These data are mixed regarding the success of the intervention on increasing breakfast calories.

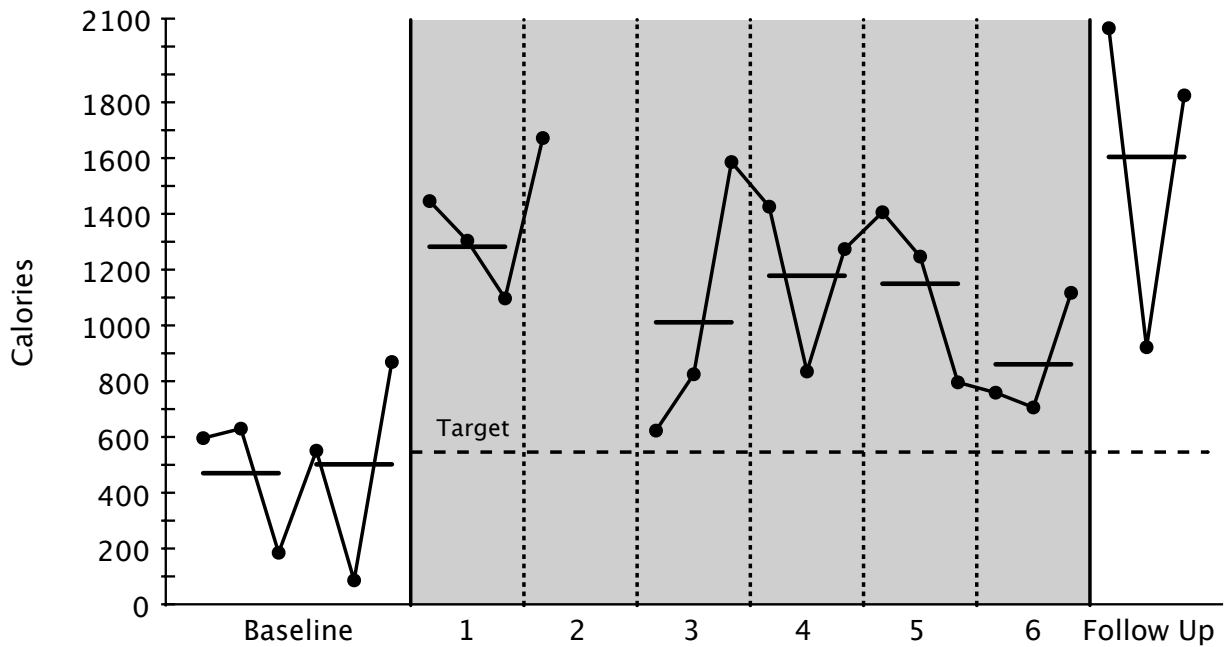


Figure 22. Chris: Total calories consumed at snack. The shaded areas connote the time during which Chris was meant to consume the target number of calories (dashed line). Short solid horizontal lines indicate weekly means.

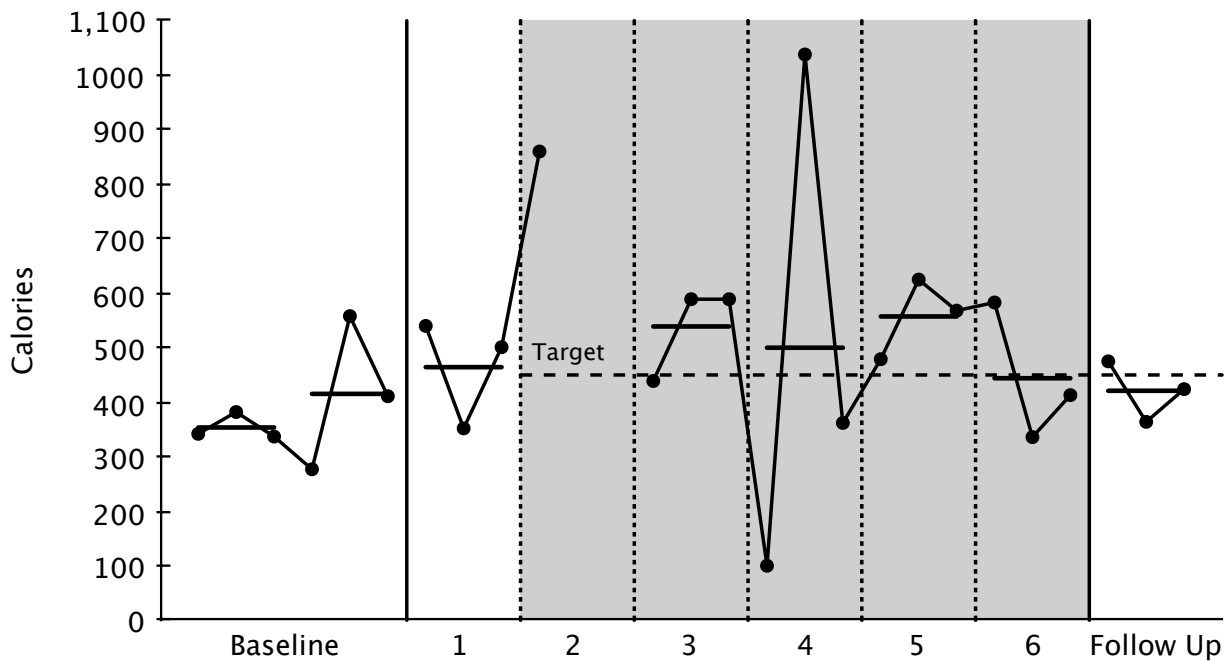


Figure 23. Chris: Total calories at breakfast. The shaded areas connote the time during which Chris was meant to consume the target number of calories (dashed line). Short solid horizontal lines indicate weekly means.

Chris’s intake at lunch is depicted in Figure 24. His intake during the first week lunch was targeted increased significantly and exceeded the target intake; however, his average intake fell during the final two weeks of the intervention; there is not a clear upward trend. During the follow-up week, his lunch intake was above the target intake levels. Overall, the data are mixed regarding the effectiveness of the intervention on increasing Chris’s lunch calories.

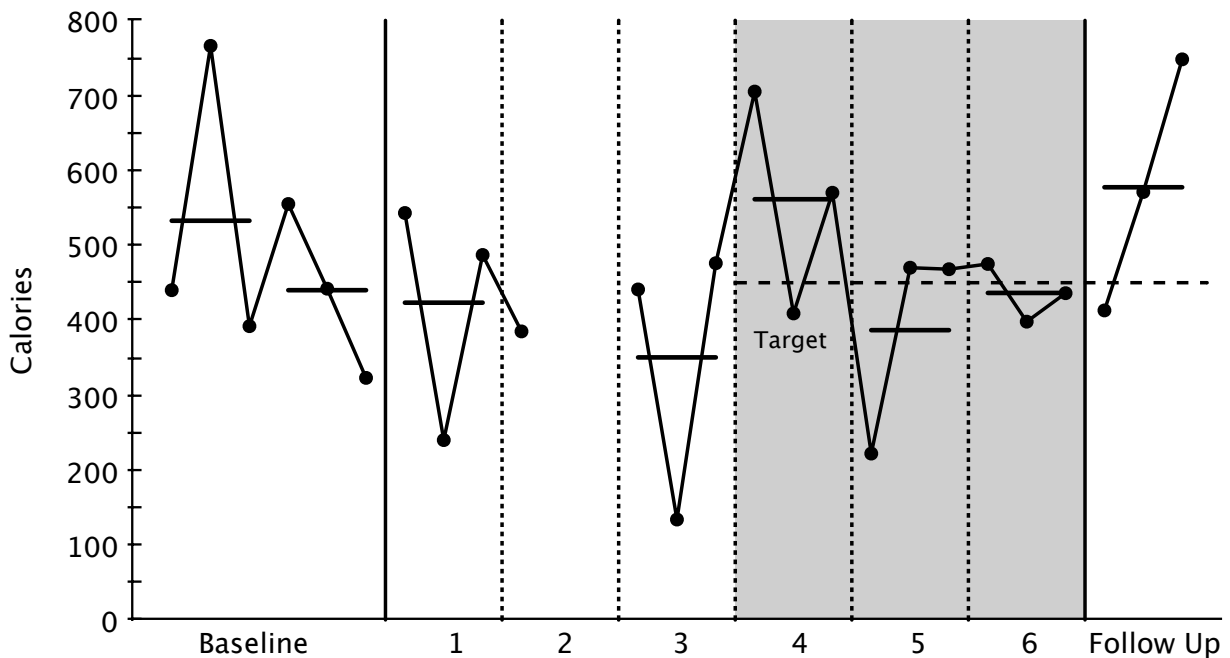


Figure 24. Chris: Total calories consumed at lunch. The shaded areas connote the time during which Chris was meant to consume the target number of calories (dashed line). Short solid horizontal lines indicate weekly means.

Chris’s intake at dinner is displayed in Figure 25. Throughout the baseline and intervention period, Chris’s average dinner intake varied and remained below the target intake levels. Although it appears there is a slight upward trend beginning in week three, a review of the data from the beginning of the intervention period indicates that Chris’s dinner intake is highly variable. Overall, the data do not suggest that the intervention resulted in an increase in calories at dinner.

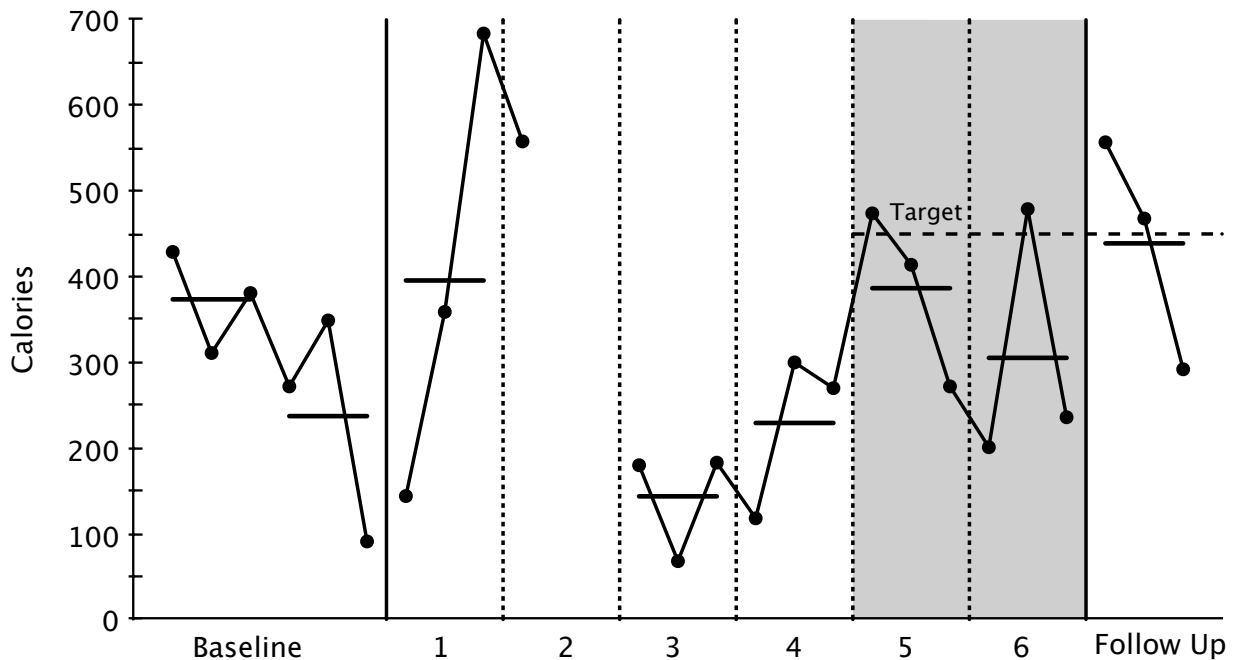


Figure 25. Chris: Total calories consumed at dinner. The shaded areas connote the time during which Chris was meant to consume the target number of calories (dashed line). Short solid horizontal lines indicate weekly means.

Secondary Outcome: Child Behavior

Results of the videotape analysis of Chris’s behavior at dinner are presented in Figure 26. These behaviors were selected as they represent behaviors that are associated with negative affect, are incompatible with eating, and are responsive to parent behavior strategies. They included: refusing to eat or complaining about food (*Refuse*), crying with or without tears (*Cry*), or getting down and walking away from the table (*Away*). During the first week of baseline, Chris demonstrated the highest levels of *crying* and *away* behavior. He also demonstrated the second highest levels of *refusal* behavior. All of these behaviors demonstrate a notable downward trend, with all behaviors occurring less frequently than once every three minutes at session six and follow-up. Overall, these data indicate that the frequency of behaviors associated with negative child affect and food refusal diminished over the course of the intervention.

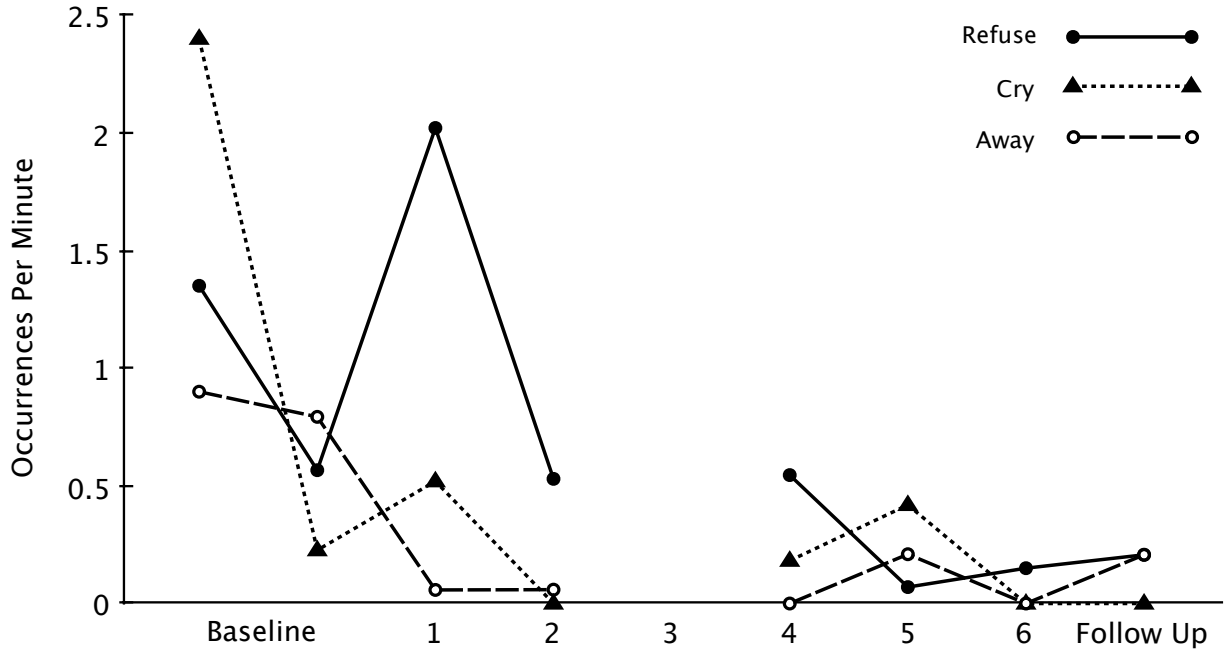


Figure 26. Chris: Rate of negative child behavior.

Overall, results for Participant 3 offered mostly positive support for the efficacy of the intervention. The primary outcome, Chris’s anthropometric data, showed clinically significant improvement consistent with the onset of the intervention. Further, these improvements in stature continued beyond the termination of the intervention, suggesting maintenance of treatment effects. Further, Chris’s overall intake increased significantly during treatment, although these increases did not occur evenly across all meals. Finally, positive effects were noted with regard to Chris’s negative affect and behavior during meals across the intervention.

Outcome Results: Participant 4, Group #2: Peter

Peter was a 21-month-old boy at the start of the intervention. His parents were married. His mother attended all meetings alone and was the primary caregiver. Peter had two older siblings living in the home. His mother had steamed, pureed, and froze a variety of organic fruit and vegetables since he was able to consume solid food, and the majority of his food consisted of

these pureed foods. His therapeutic goal was to make his meals developmentally appropriate by decreasing the proportion of his food offered in a soft, pureed texture.

Primary Outcome: Diet Evaluation

Results of Peter's diet texture evaluation are presented in Figure 27. Peter's mother cooked, pureed and spoon-fed all of Peter's food, a practice she had engaged in since he was old enough to begin solid food. Percent of food served pureed was calculated by tallying the number of items served pureed or soft (i.e., yogurt, pureed vegetables, rice cereal) and dividing it by the total number of items offered. Beverages were excluded from this calculation. During the baseline period, between 70-100% of the food offered to Peter was pureed. There is a clear downward trend throughout the weeks of the intervention, with only 0-10% of foods in the final week of the intervention meeting this definition. During the follow-up week, between 0 and 20% of the foods offered to Peter were soft in texture.

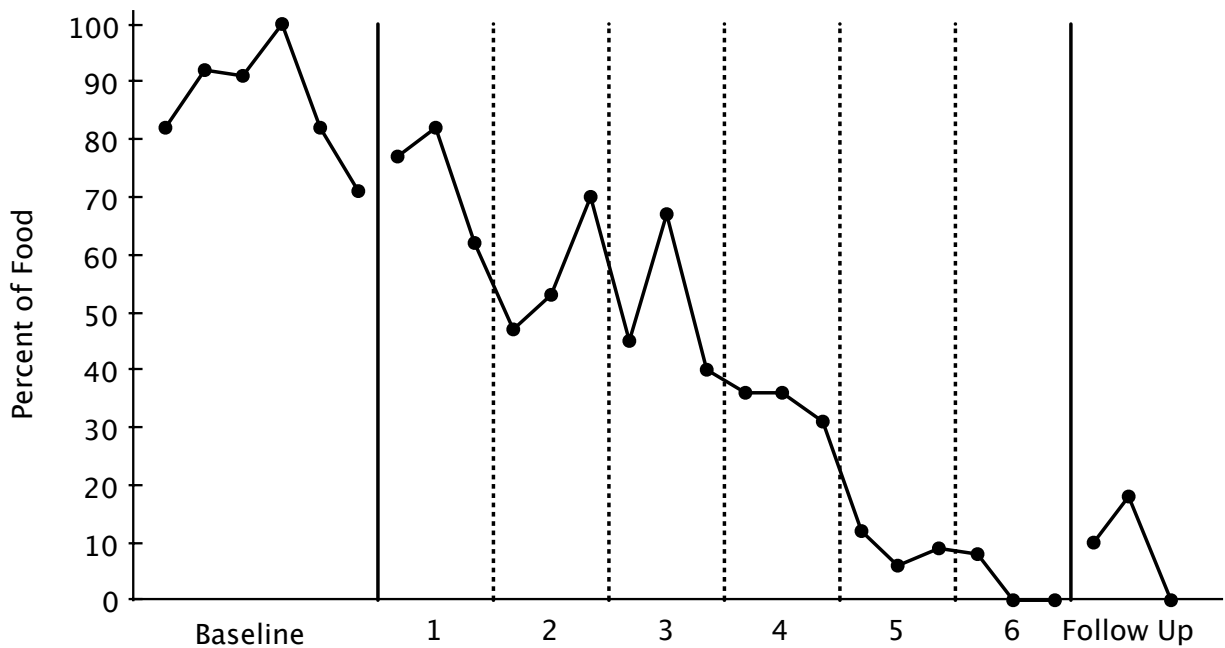


Figure 27. Peter: Percent of spoon-fed food in diet.

Secondary Outcome: Caloric Intake

Because Peter's weight to length percentile placed him in the overweight range, his treatment goals did not involve a change in caloric intake. However, his intake was tracked over the course of the intervention. His total daily intake is presented in Figure 28. His overall intake remained consistent from baseline through the intervention and follow-up periods. Overall, these data indicate that his overall calorie intake did not diminish with the introduction of developmentally appropriate foods and textures.

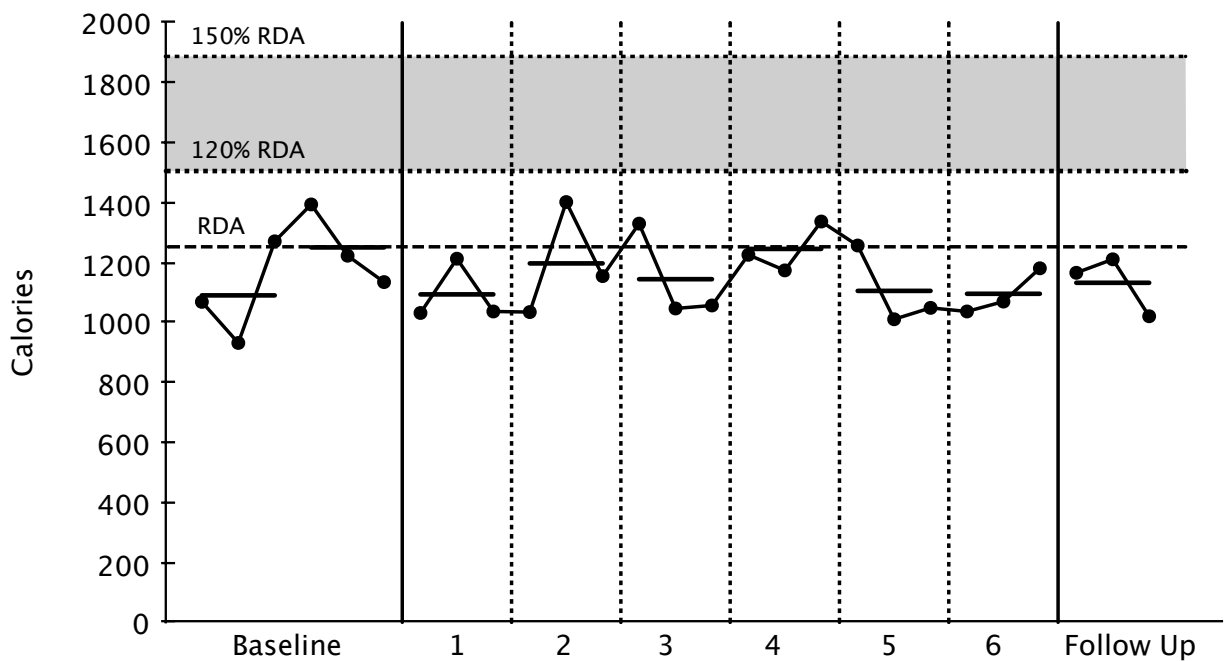


Figure 28. Peter: Total caloric intake across all phases of the intervention. Shaded area indicates CFF target range between 120% and 150% RDA. Short solid horizontal lines indicate weekly means.

Secondary Outcome: Anthropometric

Figure 29 displays Peter's body composition for the four months prior to baseline through three months after the follow-up week; Figures 30 and 31 provide Peter's length and weight on standardized curves. Throughout the months prior to the study, Peter's body composition was

consistently at or above the 85th percentile. His family was not able to provide anthropometric data for the follow-up week, so the follow-up data point was estimated. A review of Peter's growth and weight gain on standardized curves suggests fairly stable trajectories and indicates the drop in Peter's BMI percentile may be explained by the combination of a growth spurt and weight loss.

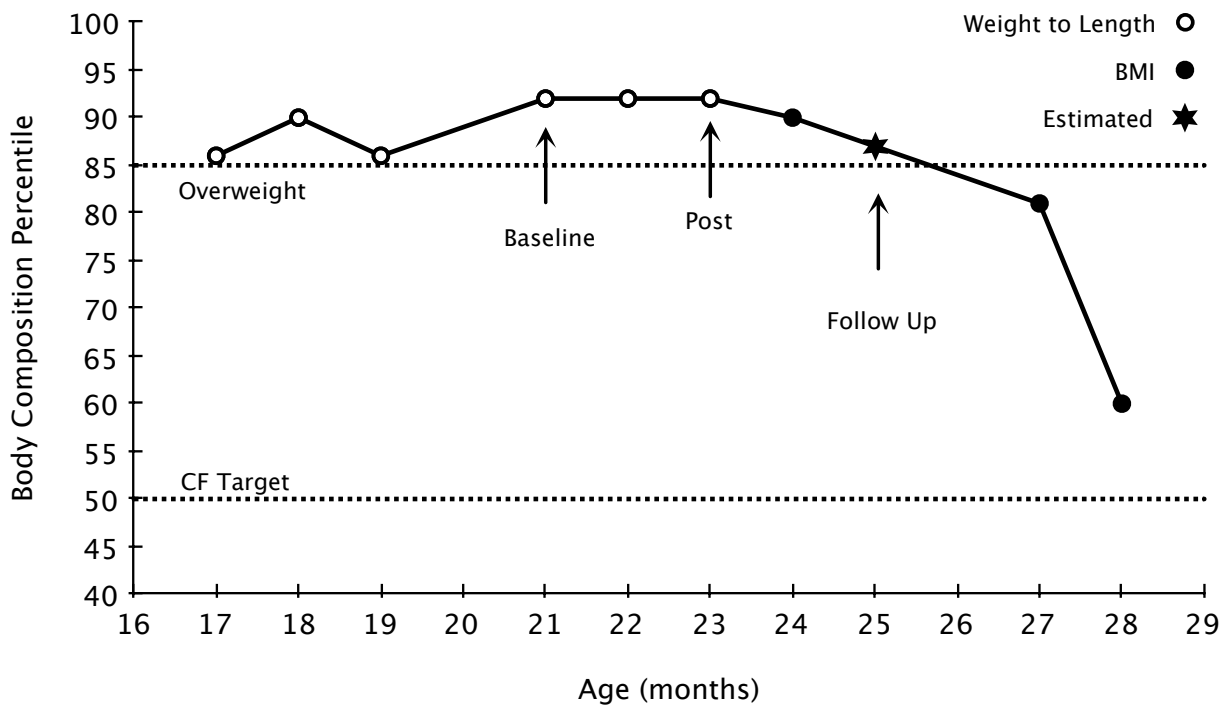


Figure 29. Peter: Body composition percentile.

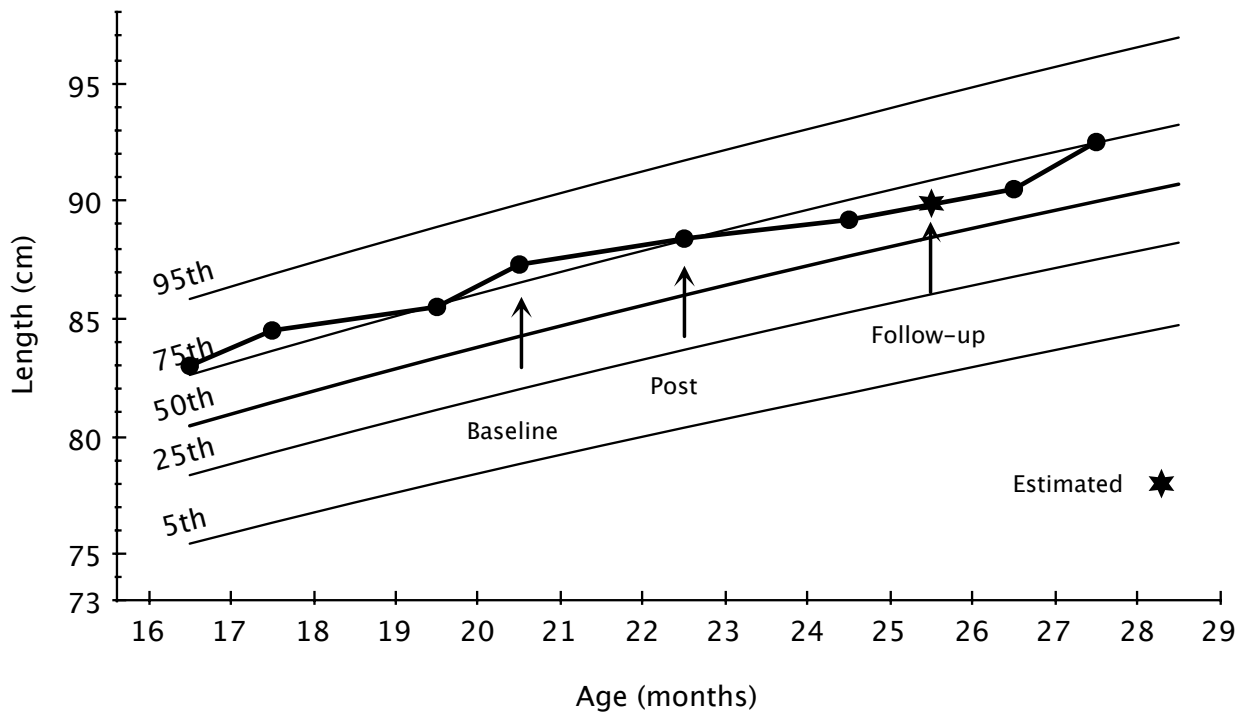


Figure 30. Peter: Length for age.

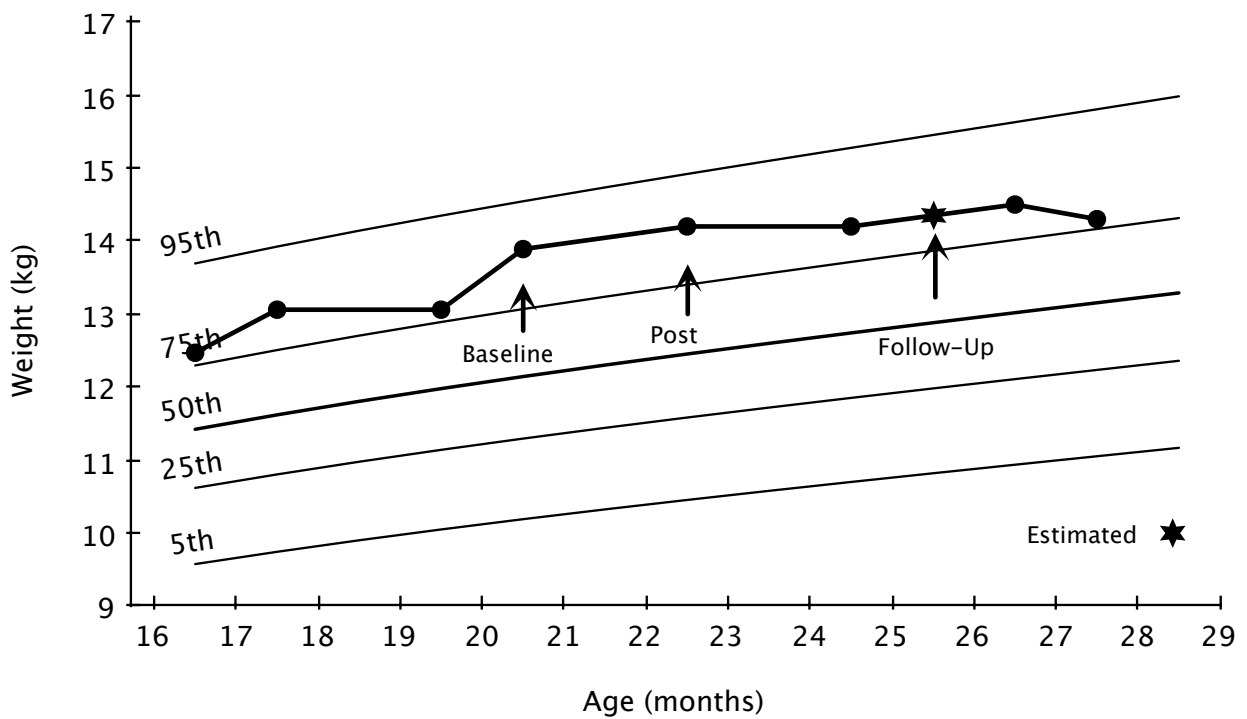


Figure 31. Peter: Weight for age.

Overall, results for Participant 4 offered support for the efficacy of the intervention. Significant improvements were made in the primary outcome, age appropriate diet. Further, these changes did not impact Peter's overall caloric intake, and his BMI percentile remained relatively stable during and after the follow-up period, although he experienced a subsequent decline.

Discussion

This study tested a developmentally-sensitive adaptation of an evidence-based treatment to increase caloric intake in toddler and preschool children with CF. Parents of four children with CF participated in the eight-week long intervention and 12-week post intervention follow up. Participants provided three-day diet diaries and one video recorded one meal each week. Anthropometric data were also collected at baseline, post-treatment, and follow-up. Efficacy of the intervention was judged for each participant separately, based on his/her individualized behavior and nutrition goals. Overall, this study provided support for the intervention for some but not all participants. Fully 75% of participants demonstrated positive change for their primary outcomes. Results for secondary outcomes were somewhat more mixed, with 75% of participants demonstrating positive change on at least one secondary outcome, with 38% of secondary measures demonstrating no change. Anthropometrics (i.e., BMI percentile, height, weight) were primary outcomes for Ben and Chris. Anna and Peter's primary outcome measures were diet evaluations based on individualized treatment goals. Secondary outcomes included caloric intake (i.e., increase intake for Ben and Chris, maintain or slightly decrease intake for Anna and Peter), parent report of mealtime behavior problems, and observational assessments of parent and child behavior.

Results suggested three of four participants demonstrated notable improvements after completing the intervention. On the positive side, two participants, Ben and Chris, who began the intervention underweight (i.e., below the 50th percentile for body composition), both gained weight during the intervention and continued these gains through the 12-week follow-up period. Peter, a child whose body composition was in the acceptable range, was able to successfully transition away from spoon-fed purees to foods with developmentally appropriate textures. On the negative side, Anna, a child whose diet consisted of highly processed, low nutrient foods did not replace these foods with more nutritious alternatives. This section will review the main findings in terms of effectiveness toward the goals of weight gain, increasing caloric intake, non-caloric nutritional interventions, and child behavior management skills in more detail, identify future directions for research and practice, then discuss limitations.

Weight gain. A disproportionate amount of children with CF are underweight (Stallings et al., 2008), a condition associated with a variety of untoward pulmonary outcomes (Lai & Farrell, 2008) and evidence suggests early body composition is closely associated with later health (Konstan et al., 2003; Lai et al., 2009). Given the research suggesting a strong protective effect associated with having a BMI above the 50th percentile for age (CFF 2007; Stallings et al., 2008), weight gain was a primary goal of this intervention and the original intervention (Stark, 2003a) on which it was based.

Two of the four participants had weight gain as their primary therapeutic goal. Both children had consistently low body composition percentiles; Ben's was below the 30th and Chris's was below the 5th, and both children demonstrated improvements in BMI percentiles beginning at the baseline time point. Further, for Chris, whose data was available beyond the follow-up, evidence suggests that treatment gains were maintained. These data provide strong

support of the efficacy of this treatment for improving weight in underweight toddlers with CF. The Be In CHARGE treatment (Stark, 2003a) was originally designed to increase weight in children with CF, and these results support the use of a developmentally sensitive adaptation of this manual to parents of toddlers and preschoolers, albeit with only two participants.

Caloric intake. The CFF recommends that all children with CF consume at least 120% of their RDA (Stallings et al., 2008). However, research demonstrates that many preschool (Powers et al., 2002; Stark et al., 1995) and school aged children (Stark et al., 1997; Tomezsko et al., 1992) with CF do not meet this requirement. Of the four children in this study, two had average intakes below 100% of the RDA across the two weeks of baseline. However, of these two children, only one was underweight. Chris’s average consumption across the two baseline weeks was above 120% of his RDA, although he was the most severely underweight child in the study and increase in caloric intake remained an important treatment goal.

These paradoxical findings illustrate the complexity of achieving energy balance in children with CF and suggest that the advice to “add more calories” is not effective for all children. The current intervention combined evaluations of each child’s unique diet with individualized recommendations tailored to each child’s tastes and preferences. The individualized review of each child’s diet across the baseline period also served to identify specific weaknesses in children's diets. For example, as a result of the close monitoring of his intake and pancreatic enzyme dosing, Chris’s mother changed the timing of the enzyme dose from midway through his meal, resulting in malabsorption of the available fats, to the beginning of his meal, which resulted in his receiving the maximum benefit from his enzymes.

Of note, increases in children’s total daily intake did not occur evenly across meals in the manner prescribed by the intervention. This discrepancy between the gradual increases across

meals expected based on previous research (Stark et al., 1993) and patterns demonstrated in Ben's and Chris's intake might be related to a variety of factors. First, the original intervention was designed for school-aged children who were also involved in the intervention and had incentive to meet their calorie goals. Toddler eating is much more variable than the eating patterns of older children (Satter, 2000) and as such, efforts to increase toddler intake may produce different patterns of results. Second, individual child or family food preferences might also be involved; for example, neither Ben's at breakfast nor Ben's or Chris's intake during dinner approached the target amounts; instead, much of both boys' intake increases were attributable to additional snacks. This is consistent with research on toddlers and preschoolers with CF increased opportunities for intake are associated with achieving calorie goals (Powers et al., 2003).

Other dietary modifications. This intervention sought to modify the diets of two children who were not at risk for malnutrition, representing a departure from the original focus of the intervention, with mixed results. Two of the four participants had adequate to overweight body compositions but consumed diets that were considered deleterious due to either the nutritional quality (Anna) or the highly restricted consistency (Peter). As such, the scope of the intervention was extended to address a broader range of pediatric feeding problems, including those associated with pediatric obesity (Phares, Curley & Rojas, 2008) and texture acceptance (Linscheid et al., 2003).

Anna's case demonstrates that among children with pediatric chronic illnesses that increase risk for failure to thrive, obesity may still be a significant health risk. Further, these children may be at risk for nutrient deficiency due to the combination of not receiving many nutrients (Gillis & Gillis, 2005) and problems with absorption (Durie & Pencharz, 1989).

Research focused on improving children's diets reveals the complexity of the influences that contribute to pediatric overweight and obesity (Phares et al., 2008; Vereecken, Keukelier, & Maes, 2004), and family-based interventions often incorporate parental weight-management goals to promote healthy habits that benefit all family members (Jelalian & Mehlenbeck, 2003). However, the emphasis on high-calorie diets and weight gain in the treatment of CF provides an additional layer of complexity to the treatment of children with CF and overweight. In Anna's case, her parents employed many coercive and authoritarian strategies associated with increased feeding problems and disruptive behavior (Patrick, Nicklas, Hughes, & Morales, 2005; Sanders, Patel, le Grice, & Shepherd, 1993) to encourage her intake of highly processed low nutrient food.

The dietary changes prescribed for Anna were consistent with CDC recommendations to increase consumption of fruit and vegetables and reduce intake of sugary beverages to reduce the risk of pediatric obesity (Sherry, 2005), however, the data indicate that the intervention demonstrated minor, if any, success in these two endeavors, as Anna's diet retained a low level of poor quality food even as her caloric intake decreased. There are a number of possible reasons for this result. First, it may be that Anna's diet reflected the eating habits and diet of the entire family, therefore changes in her diet to increase healthful foods would likely have required changes to the whole family (Jelalian & Mehlenbeck, 2003; Phares, Curley, & Rojas, 2008). Second, whereas this may have been possible, these efforts may have been impeded by Anna's parents' belief that their current approach was effective, as Anna's weight was consistently high and she did not have the concerns about low body weight expressed by Ben's parents. In this way, having this mixed group may have been less effective as the educational needs of the families were different, and the structure of the intervention may not be ideally suited to addressing mixed treatment goals.

Unlike Anna, Peter's diet was of generally high nutritional quality. Since he achieved the ability to eat solid food, his mother had chosen to steam, puree, and spoon-feed him a variety of organic foods. This practice provided high quality nutrients, but restricted his development of feeding skills. For example, Peter did not have the opportunity to explore a variety of foods, consume a variety of textures, develop preferences for greater texture complexity, or experience the range of flavors consumed by other members of his family (Linscheid et al., 2003). Further, these restrictions may have implications for development of his feeding skills. Research has demonstrated that highly restrictive parent-led feeding may contribute to poor control of appetite and weight management later in life (Costanzo & Woody, 1985), including responsiveness to hunger and satiety cues (Birch, 1990). It is also possible that this practice conferred some benefit insofar as providing protected one-on-one time during which his mother was able to provide contingent reinforcement within the spoon-feeding practice. Further, the oversight of his eating may have served to reassure her that he was eating. As gaining weight was not a treatment goal, Peter's mother was not provided with specific instruction to increase his caloric intake as a target of the intervention. However, she was provided with all of the information about how to increase calories in the future and was encouraged to provide Peter with calorie dense and developmentally appropriate foods.

Child behavior management skills. This intervention sought to provide parents with the child behavior management skills to facilitate good eating behavior. Research has demonstrated that toddlers with CF behave nearly identically to toddlers without CF in regard to their eating behavior, but parents of children with CF often perceive their children as having more frequent and severe behavioral problems at mealtime and endorse using strategies involving coercing or cajoling to encourage their children to eat (Powers et al., 2002). These strategies inadvertently

reward children's refusal or not eating through the provision of parental attention, but parents' behavior is intermittently reinforced by the child's eating, which then leads to increased use of this parental strategy (Sanders et al., 1993). This maladaptive reciprocal pattern of mealtime behaviors is theorized to begin in early childhood and escalate throughout childhood (Bowen & Stark, 1990; Stark et al., 1995), contributing to lower child weight, poorer child health, and increased familial stress. The goal of this and other similar interventions is to provide anticipatory guidance to families during a developmental period when children's eating behavior is marked with significant changes, including refusal to eat, avoidance of previously enjoyed foods, and significant variability in intake from one day to the next (Satter, 2000).

The data were somewhat mixed regarding the effectiveness of the intervention for changing parental behaviors. The video data were not as applicable to the original aims of the study (i.e., measuring change in parent behavior throughout the course of the intervention) as was originally hoped for a number of reasons. First, the nature of in-home data collection lends itself to a variety of interfering artifacts (i.e., siblings) that added a great deal of noise to the data. Second, several parent participants disliked being on camera and either positioned the camera so they were completely out of frame or, occasionally, failed to capture the entire meal. Although naturalistic observation was important for one of the goals of the study (i.e., to improve toddler eating at home), the data as collected had limitations for the detection of behavioral change.

Further, given the transactional nature of parent-child interactions (Sanders et al., 1993), changes in parent behavior that result in improvements in child behavior may be difficult to detect using observational coding. However, there appeared to be some positive changes. Chris's negative affect at the video recorded meals also declined over the course of the intervention; this may have been due to chance or a return to baseline after spuriously high levels of affect during

baseline. Alternately, this decrease may also have been related to changes in his mother's behavior or to decreases in pain and discomfort secondary to incorrectly administered enzymes.

As Ben's weight and intake increased, his mother's report of problem behaviors on self-report measures fell. Her change in perception might reflect a decrease in conflict concurrent with an increase in her use of the child behavior management strategies taught in the intervention. It might also reflect a decrease in her stress around his eating. Her report of his behavior on the BPFAS was consistent with other parent reports of toddlers. The sub-clinical frequency scores, indicative of toddler eating behavior within normal limits, and clinical problem scores, indicative of parent perception of problematic child behavior, was consistent with other research of parents of toddlers and preschoolers with CF suggesting that at a young age, children with CF behave similarly children without CF, but parents of children with CF perceive much greater problems in children's eating than do parents of non-chronically ill children (Powers et al., 2002; Stark et al., 1995). Interestingly, Ben's father did not perceive the same problems, and his report remained below the clinical levels.

Limitations

There are several limitations to consider with regard to this study including small sample size, limited external validity, and limitations in assessment. Given the small population from which the sample was drawn, parents of toddlers and preschoolers who were patients of the CF clinic, the sample size was limited from the start. Further, it is possible that families who elected to participate were somehow distinct from those who declined participation, introducing the possibility of selection bias. Although approximately one third of the population of interest participated, the small sample size limited variability within the sample and as such generalizability to a wider population of parents of toddlers and preschoolers with CF. Given that

all participants were drawn from a relatively small population of families living in a specific geographic area and who attended the same CF clinic for their medical care, future research drawing subjects from a wider population is necessary to provide additional support to the effectiveness of this intervention.

Second, the question regarding mechanisms of change remains unanswered. Linking mechanisms of change to a clear conceptual framework facilitates the advancement of programmatic research to improve identified outcomes (Drotar, 2002). The limited video behavior data restricted the inferences that could be made about parents' uptake of child behavior management skills. In the future, researchers might consider using a research assistant to collect *in situ* behavioral video data at baseline, midway through the intervention, post, and follow up. Use of a research assistant would reduce parental data collection burden and facilitate adherence to video recording procedures.

The issues of generalizability and portability present an additional limitation. In the current investigation, the treatment was delivered by a clinical psychology doctoral student with advanced training in evidence-based treatment delivery, cognitive-behavioral case conceptualization, and familiarity with CF. During the first group, the clinic's RD participated in the first session to help the PI answer questions about dietary monitoring. During the second group, a second doctoral student with training similar to the PI participated in treatment meetings to facilitate data collection. A few points are relevant to study limitations here. First, whereas the present findings indicate this intervention may be delivered without a doctoral level clinician, very few CF clinics in the country have access to clinical psychology doctoral students. Thus, more research is needed to determine whether similar outcomes could be achieved if other professional members of the CF team delivered the treatment (e.g., social workers, registered

dietitians, nurse practitioners). Second, that the RD was involved in one session for group 1 but none for group 2 could make interpretation of these results a challenge. Although the RD was not physically present for the sessions of the second group, her involvement in the planning of nutritional recommendations was equivalent across both groups, a fact that mitigates her physical absence from group 2.

Future Directions

The current study suggests multiple future avenues for research related to improving the nutrition of young children with CF, including expanding the scope to evaluate generalizability, exploring options for shorter, more time-intensive formats for treatment delivery, and investigating the utility of video feedback to enhance training. First, future research might expand upon the current intervention to involve larger samples, including the involvement of participants from other CF clinics, the use of wait-list control groups or a randomized clinical trial. The involvement of additional CF centers or additional research designs would provide additional evidence for the efficacy of intervention. Further, researchers might consider altering the procedures for behavioral data collection to facilitate the collection of high-quality video data. For example, employing research assistants to ensure high-quality naturalistic video data or including video recorded meals in the lab might provide a clearer picture of behavior at each time point and facilitate the evaluation of the hypothesized mechanisms of change.

Second, Annie and Peter's data indicate that children with CF with high body composition percentiles who may not be identified as requiring nutritional guidance may have significant nutritional or behavioral concerns. Future research exploring the effectiveness of combined group behavioral interventions with groups separated by nutrition status is needed to determine the most effective method of intervention delivery to parents of toddlers and

preschoolers with different nutritional needs. Previous intervention research has identified and targeted children based on their body composition; however, it is possible that this intervention is effective for a broader range of children. Examining both dietary and behavioral needs as a predictor of treatment response may provide insight into which families may be most likely to benefit from this or a similar intervention.

Third, several parents who declined to participate cited time constraints and not having current behavioral concerns as their reasons for declining. Parents of children with CF have significant demands on their time associated with CF treatments, including myriad medical visits. However, lessons learned from this intervention indicate that BMI percentile is not a reliable indicator of quality of a child's diet. Identifying a shorter, possibly more intensive format for the intervention might provide options for increasing the access for caregivers. It might also be valuable to consider widening the age range for parents of older children with behavioral eating concerns. Some clinics provide annual weekend workshops with topical speakers and short courses for families; providing didactic sessions across one or two weekends might increase access for families and increase interest in and awareness of the availability of behavioral interventions.

Finally, future research might explore the use of video in providing direct and targeted feedback to improve acquisition of skills taught in the intervention (Hosford, 1980). Interventions using combined group and individual formats to improve toddler behavior have utilized video review of skills taught in the group session to provide individualized strengths-based feedback to parents (Sharry, Guerin, Griffin, & Drumm, 2005). Particularly given the widely variable nature of the individual treatment goals for parents, a combining the group sessions with individualized video-feedback sessions may provide both the opportunities for

individualized critiques without violating confidentiality (i.e., by sharing the video with the group). This might also provide the opportunity to address concerns parents are not willing to identify in a group format.

Practice recommendations. In addition to future research, the current study suggests several recommendations for implementation of this treatment in a medical center context. Before discussing those recommendations directly, it is important to make a note about the generalizability of the behavior management strategies used in this intervention. These strategies, including differential reinforcement of other behavior, reward and reinforcement, and establishing rules and consequences, are integral in the treatment of many problems of childhood, including oppositional, defiant, and aggressive behaviors (Kazdin, 2005), attention deficit hyperactivity disorder (Barkley, 2006), childhood anxiety (Chorpita, 2007), nocturnal enuresis (Houts, 2010), and pediatric encopresis (Ritterband et al., 2003), among others. Further, these principles are applicable to non-clinical populations and are often taught to parents as helpful general child behavior management skills. Although the nutrition recommendations in the current study are specific to children with CF, the behavioral components are generalizable to a broad range of illness specific and general parenting concerns, such as adherence to a variety of medical treatments as well as effective discipline and management of tantrums. As such, the applicability and usefulness of this intervention may extend beyond diet change to other pediatric concerns (i.e., treatment adherence), specifically if generalization of child management skills to these behaviors are explicitly addressed and discussed within the intervention.

This study also suggests that children who might not otherwise be identified as having difficulty with feeding due to their above average BMI percentiles may in fact have aberrant behavior that may require intervention. Given the importance of feeding and nutrition in CF care,

providers might consider using the BPFAS as a routine screening tool to identify potential problems and provide timely intervention. Further, given the change in eating behavior over the course of child development, families who receive intervention should be monitored and offered the opportunity for additional “booster sessions” to facilitate the maintenance of treatment gains. Finally, providers might benefit from training in motivational interviewing skills (Rollnick, Miller, & Butler, 2008) to enhance non-confrontational and non-adversarial communication with patients and families to facilitate a collaborative relationships and elicit motivation for health behavior change.

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Appendix A

Recipe Form

Name of Ingredient	Amount In Recipe	<i>Calories in Recipe*</i>
<i>Total:</i>		

What is the total weight after cooking?

-or-

How many servings?
What does each weigh after cooking?

Please feel free to attach any nutrition labels to the recipe.

**Calories in recipe are optional.*

Appendix B

DINE CODE

General Coding Guidelines

- 1) In general, the unit of verbalization coded is the phrase. A phrase may consist of a string of words or a single word. Whether or not a particular word or phrase is scorable is dependent upon the criteria that define the individual coding categories. Pauses (defined grammatically or by time) between phrases define the onset of a new verbalization. Additional information regarding what constitutes a scorable verbalization is presented within the description of specific codes.
- 2) When assigning a code to a behavior, a 2-step process should be followed. The first step is to identify the content of the verbalization and assign it to a coding category. When there is ambiguity about appropriate assignment based solely on content, the second step should be to utilize the context in which the behavior occurred. While this process may not always eliminate the ambiguity, it should provide further information for assignment on the basis of the definitions in the manual. It is important to remember that content is the primary basis for assignment to a coding category.
- 3) A particular behavior should be assigned to a coding category at the point at which the nature of the behavior becomes clear; that is, at the point at which it can be clearly identified as meeting the criteria for a particular code. Thus, if a behavior starts in one interval, but only becomes identifiable in the next, it should be coded in the second interval. This is distinguished from those situations in which a behavior is clearly identified in the first interval and overlaps briefly with the interval change. In this case, the behavior is scored in the first interval.

PARENT BEHAVIORS

Included here are behaviors emitted by the child's parents as well as other adult figures who are known to be in regular attendance at meals. Behaviors emitted by siblings are not coded as parent behaviors.

COMMAND - ALPHA (C):

Definition: Any parent verbalization specific to eating -this includes chewing, swallowing, and drinking - to which a motoric response is appropriate and feasible. Commands may be in the form of an order, suggestion, question, rule, or contingency, as defined below. Statements or questions that are in the form of commands, but inhibit eating, are not included here. **To score a parental verbalization as a command, a verb is a necessary but not sufficient component.**

Direct Commands- include orders to eat (including swallowing and chewing) that are stated directly and specify the child behavior to be initiated. Verbalizations that do not specify eating or drinking explicitly, but clearly refer to this behavior in the context, may be coded as commands. For example, "Open up", when it is clearly in reference to taking a bite of food, would be coded as a command. Choice commands are coded as commands.

Examples: "Eat your peas."
"Drink your milk or take a bite."
"Take a bite."

Indirect Commands - include suggestions to eat that can be responded to motorically, that are not in question form.

Examples: "See if you can eat some more."
"Let's take a bite."

Question Commands - include questions that direct the child to eat and to which both a motoric and a verbal response is available. To be scored as commands, questions must direct rather than follow the child's behavior. For example, "Are you eating?" would be coded as a Parent Talk rather than as a command because the question does not imply a directive for the child to eat.

Examples: "Are you going to eat your peas?"
"Will you have some more?"

Permission Statements and Rules - include statements ("You have to eat dinner") or rules (implied if/then commands without the "if...then..." structure) that specify an eating behavior to be initiated in the present.

Examples: "You should eat now."
"We are going to take a bite."

If / Then Statements and Warnings- include contingency statements in which the "then" component refers to a positive, neutral or negative consequence to be administered by the parent. Thus, the part of the phrase is the command, while the "then" component is simply information that is not coded. Also includes statements that are verbalized as "then if".

Examples: "If you eat your potatoes, you can have dessert."
"I'll be proud of you if you eat everything on your plate."
"There will be no ice cream if you don't eat your potatoes."

Chain Commands - a series of related commands about eating that are given in rapid succession without pause. In nearly all cases, the cue for scoring two or more commands as a chain is that they

are connected by a conjunction. A command is scored as long as one part of the chain refers to eating.

Examples: "Eat some peas and finish your potatoes."
"Take a bite and drink some milk."
"Sit down and drink your juice."

If there is a pause between commands in a chain, then the coding is as a series of interrupted commands. In this case, they are defined as Beta Commands.

Compliance and Noncompliance to Commands (“+” or “-“ respectively):

Definition: Any time that an Alpha Command is coded (and does not default to a Beta Command), compliance or noncompliance must be scored. The criterion for compliance is that the child initiates effort at eating (chewing, swallowing) within 5 seconds of the parental command. A child may be judged as compliant without a bite being scored - e.g. a first step toward eating, such as picking up a utensil, may be enough of an effort to be judged compliance.

The 5-second interval for assessing compliance begins at the end of the parental command. If the parent offers a command when a child is in the process of initiating or taking a bite, then effort in response to that command can be coded as long as the child has not completed taking the bite. In the case of a chain command, the 5 second interval begins at the termination of the final command in the chain. (In some cases, parents will give commands to eat a specific food - e.g. "Eat your potato." Compliance can be scored in this situation as long as the child makes an effort to eat some food. S/he does not have to eat the food designated by the parent. Noncompliance is scored if the child takes a drink rather than eating food.)

Compliance is scored independently from the child behavior category of "Refuse/Complaint". That is, a judgment regarding compliance/ noncompliance should be made separately from the child's verbal or nonverbal refusal. For example, a child may initially offer a verbal refusal in response to a command, but may then comply to the command within the 5 second interval. In this case, a refuse and compliance are both scored. The full 5 seconds should always be awaited before noncompliance is scored.

COMMAND BETA (C/)

Definition: Beta Commands are defined as those commands that would be Alpha Commands by virtue of content, but are scored as Beta because of the process by which they are communicated. These are commands that fall under one of the six categories defined above, but for which there is no opportunity for compliance. There is no opportunity for compliance under the following circumstances:

1) The command is followed (within 5 seconds) by parental verbalization before the child complies. This occurs when: a) the parent giving the command interrupts him/herself with talk to the target child or b) the parent who did not give the command directs a verbalization toward the target child. Interruptions can occur when the parent completes a command and then immediately utters another word/phrase/sentence that is grammatically separate. For example, "Take a bite. Please." would be scored as an interrupted Alpha Command (i.e. Beta Command) followed by a Coax. This is distinguished from the sentence, "Take a bite, please.", which would be scored as an uninterrupted Alpha Command. Interruptions to commands also occur any time the parent utters a word/phrase/sentence (that is not part of the command) within 5 seconds of the initial command.

Examples - Interrupted Commands

"Have a drink." (3 sec.) "Ben". "Chew your food."(no pause) "Come on." "Eat that." (no pause) "Go ahead".

Examples - Uninterrupted Commands

"Eat that, will you?" "Take a bite because you're the only one who's going to eat it. " "Have a drink, Ben."

2) The parent restricts the child's mobility or removes food/drink, thereby precluding opportunity for compliance.

Note: For dissertation coding, commands that are followed by words that may or may not be grammatically separate (i.e., "Take a bite, ok?") are not coded as beta commands by virtue of the final word and the question of whether a period or comma separated the two phrases.

The second phrase after an interrupted beta command can be coded as an alpha command (i.e., "Take a bite. (3 sec). Ben, take a bite.").

COAXING (CX)

Definition: A verbalization about food or eating directed at the target child in an effort to get him/her to eat. Includes questions and statements regarding food and eating that do not qualify as alpha commands, but that have as their goal increasing the child's consumption. Does not include parent's response to requests for food made by the child, e.g., not scored if child asks for bread and parent responds, "Oh, would you like some bread?"

Offers of Food - questions asking the child if s/he would like a particular food or a second helping of a food. This includes desert foods if offered for consumption during the meal, rather than after the meal or in the future.

Examples: "Would you like some more potatoes?"
"Do you want some milk?"
"How about some ice cream?"

Non-example: "Would you like to have ice cream later?"

This category is differentiated from that of Question Commands by virtue of the fact that offers suggest only a verbal response.

Evaluation of Food - questions or statements that encourage evaluation of the food in a positive manner. Key words indicating positive evaluation include: good, delicious, like, umm, yummy, and great.

Examples: "Isn't this hamburger good?"
"Don't you like apple juice?"
"Do you like peas, Ben?"

Non-example: "How is the hamburger?"

Coaching - encouraging child to eat by making eating a game. In this situation, all aspects of "game-playing" would be coded as a Coax, even if the behavior continued after the child took a bite.

Examples: Playing "Airplane" when feeding the child

Saying "Don't eat that!" as a game to get the child to eat

Vague Commands - these are orders or suggestions that are so vague that it is unclear that eating is the behavior to be initiated. Also includes statements that offer the child a choice about eating.

Examples: "Come on."
"Eat like a big boy."
"Eat that if you like."
"Take a bite when you sit down."
"If you want to, you can take a drink."

Sarcastic/Coercive – these are statements or suggestions that seem to be attempting to trick the child to eat using "reverse psychology." A taunting, coercive, or sarcastic tone is part of this category.

Examples: "Oh, you don't like it? Mommy loves this!"

Exceptions: None of the above behaviors is scored when it is in reference to water or to seasonings such as salt and pepper.

REINFORCEMENT (RN)

Definition: Positive verbal or physical behavior by the parent that is directed to the target child following the child's eating or drinking. This code is scored on an occurrence/non-occurrence basis within an interval.

Verbal Reinforcement - includes general positive statements and positive descriptive statements of child's eating that follow the child taking a bite or sip. Statements may specify the behavior being reinforced e.g. "You're doing a good job eating", or be more general - e.g. "Wonderful!". Also includes descriptions of the child's eating or drinking as better-than-average.

Examples: "Good job."
"I like it when you eat your peas."
"You're eating so fast."

Physical Reinforcement - includes positive physical contact such as pats, hugs, kisses, or strokes that follow the child taking a bite or sip. Also includes some physical gestures that do not involve contact, but that are clearly positive responses to child's eating.

Examples: Clapping
Thumbs up

Verbal and physical reinforcement are coded according to the following criteria:

1. *Global positive statements* (e.g. "Good job") are coded as reinforcement *only when they follow bites or sips* and occur temporally in the same or subsequent interval as a bite/sip.
2. Positive descriptive statements that are specific to eating behavior (bites, sips, or chews) are always coded as reinforcement and do not have to follow a particular bite, sip, or chew. The only qualification is that only one can be coded within a given interval.
3. There can be only one reinforcement per bite, sip, or chew. This is relevant only for physical reinforcers and global positive statements, as specific praise may be scored in subsequent intervals with or without a new bite.

4. If the same parent gives additional global praise for the same bite, sip, or chew, then the second is not coded. This does not default to talk.
5. Taking vitamins or capsules is not scored as a bite. Consequently, praise for this behavior is not regarded as reinforcement.

PARENT TALK (PT)

Definition: Verbalization that is directed from the parent to the target child that does not meet the criteria for Command, Coaxing, or Reinforcement. Includes general talk from a parent to the family as a whole. A verbalization is defined as a complete phrase or a single word that stands alone.

- Examples:
1. Conversation may include any topics.
 2. Prayer is included in this category if the parent is praying aloud.
 3. For children under the age of 3, category also includes parent vocalization of sounds directed at the target child.

PHYSICAL PROMPT (XP)

Definition: A physical action by the parent to indicate to the child to eat or drink, or a physical action that serves to assist the child in taking a bite or sip. This category does not include occasions when the parent actually places food in the child's mouth (Feed).

- Examples:
1. Parent pushes the child's plate closer to the child.
 2. Parent points at the child's food.
 3. Parent scrapes child's food into a pile.
 4. Parent takes the child's hand and places it on his/her fork.
 5. Parent holds the child's hand on the spoon or cup and brings it to the child's mouth.
 6. Parent brings cup to child's mouth without child's assistance.
 7. Parent places food on utensil and then puts it down on child's plate without attempting to feed.

Guidelines:

1. Other parent behaviors, such as coax or command can be coded with prompt if they occur simultaneously. For example, if the parent points to the rice and says "Eat your rice.", both prompt and command would be coded.
2. Cutting the child's food is not considered a prompt.
3. Correcting the child by removing the child's hand from the plate (i.e., if child is playing with food with hands while utensils are available) is not considered a prompt.
4. The initial placement of the plate in front of the child at the beginning of the meal is not coded as a prompt. In addition, presentation of new food (and accompanying condiments) at any point during the meal is excluded from the category.
5. Additional servings of already presented food placed in front of the child are considered a prompt regardless of whether the child requests the food or not.

EATING BEHAVIORS

BITE (B)

Definition: Any taking of solid food into the mouth. The food must pass between the child's lips before it is considered a bite. Unlike a feed by the parent, movement of the food toward the mouth is not considered sufficient. Only code a bite after the food passes the child's lips. Taking vitamins, capsules, or other forms of medicine is not counted as a bite.

Examples of 1 bite:

1. Child takes multiple bites of corn off the cob WITHOUT removing the cob from his/her mouth.
2. Child requires more than 1 interval to suck spaghetti into his/her mouth.
3. If the child uses his/her thumb and index finger to pick something up from plate or table and places it in his/her mouth, regardless of whether the food is obvious.
4. If food falls out of the child's mouth, but is immediately replaced within the current interval, it is coded as one bite not two. In addition, a "Spit up" is not coded since the food fell out and was replaced. If the child eats what has fallen out in subsequent intervals, then it is coded as a new bite as appropriate.

Examples of behavior NOT to be scored bite:

1. Putting an empty utensil in the mouth.
2. Taking a filled utensil from a serving dish to the plate, where food is removed, after which the serving utensil is brought to the mouth.
3. Licks: licking fingers is not coded as a bite unless there is an obvious glump of food on fingers.

SIP (S):

Definition: Bringing a glass, cup, straw, or spoonful of liquid to the lips. Includes water. A sip is terminated by removal of the vessel from the lips.

Drinking from bottle: When an infant is bottle-feeding, continuous drinking may be coded. In the case of continuous bottle-feeding, a sip is coded in the interval in which the sip begins. An arrow is started in that interval and drawn continuously down through intervals until the sip is terminated by removal of the vessel from the lips. If a new sip or continuous drinking resumes, a new sip is recorded next to the "S" and an arrow is started and continued through the intervals until the sip is terminated.

Drinking from cup: When an infant or child is drinking from a cup, only discrete sips can be coded. If the child drinks continuously from a cup, this is represented by coding 1 sip in the interval in which the child begins to drink. For a new sip to be coded, the cup/glass must be removed and then brought back up to the lips. There is no limit to the number of discrete sips that can be scored within an interval.

Note: If child is drinking from a "sippy cup" and using it like a bottle (i.e. drinking for more than two intervals consecutively), then code as if child were drinking from a bottle (with continuous arrow for the length of "sip")

SPIT-UP (Sp):

Definition: Refers to any time the child spits-up, spits out or purposely drops food from his/her mouth. Action may be deliberate or involuntary. Also included are instances when food

comes out of the mouth as a result of overfeeding and is not replaced. A BITE MUST BE CODED BEFORE A SPIT-UP CAN OCCUR. if the child purposely removes food from his/her mouth (with hand or utensil) and then replaces it within the same or subsequent interval, this is not coded as spit up. The normal mess of food on the mouth during infant feeding is not considered "spit up".

FEED (F)

Definition: Any time the parent, other adults, or siblings put or attempt to put food in the child's mouth with no attempt to have the child help. Includes feeding of solid foods as well as bottle feeding. **However, when** liquids are presented in a cup rather than a bottle, the scoring is Physical Prompt instead of Feed.

Children 3 Years and Older

Onset of a Feed: For this group, onset of a feed begins when the parent starts toward the child's mouth with the food. (i.e. the motion of the parent's hand indicates the onset of a feed.)

Feed Without a Bite: If the parent brings the utensil or food back to the plate or down to a resting position without putting food in child's mouth, then a feed without a bite is scored. if the parent then successfully re-feeds the child, a new feed and bite is scored.

Children 2 Years and Younger

Onset of a Feed: The onset of a feed occurs when the food is presented within 3 inches of the child's mouth. (We are trying to differentiate between an actual feed and the parent simply getting the spoonful ready.)

SCORING:

1. Feeds and Bites are usually scored in the interval in which the bite takes place. For example, if the feed occurs in 1.2 interval and the interval changes while the food is going into the mouth and it passes the lips in 1.3, score both the feed and the bite in 1.3.
2. If the food is held within three inches of the mouth until the child accepts the bite, score both a feed and a bite in the same interval in which the bite occurred.
3. A feed would be scored without a bite if (1) the child refuses (see definition of refuse) (2) if the parent withdraws the spoon after presenting it within one inch of the child's mouth.
4. If the parent withdraws the food out of the 3" range and then goes back within the 3" range and there is a bite, the original presentation is a feed without a bite and the second is a new feed and is coded with the bite.

PLATE AWAY (NP)

Definition: Any time someone other than the target child moves the child's plate out of his/her reach, such that the child does not have opportunity to take a bite. Plate must be removed and kept away from the child for an entire interval to be coded. Solid food must be presented to the target child and then taken away before a NP can be coded (i.e. NP is not coded if child begins meal without a plate and with a bottle). If the child takes his/her own plate to be refilled, this is coded as "CHILD AWAY" rather than "PLATE AWAY" (See definition of Away under Child Behaviors).

Examples: Parent removes plate to add more food.

Nonexamples: Target child pushes plate away.

Target child stands in chair and is out of reach of plate. (This would be coded "Away" if

under Child Behaviors.)

Parent moves plate in front of self to feed child.

Child drinks from bottle/sippy cup before food is initially presented
Plate is removed, but other food source is present.
End of meal.

CHILD BEHAVIORS

REFUSE FOOD/COMPLAINTS ABOUT FOOD (R):

Definition: Any time the child lets the parent know that he/she does not want more food or complains about the taste of food. Refusals may be verbal or non-verbal. Complaints are by definition verbal and refer to any negative evaluation of the taste, flavor, or texture of food. Complaints about the temperature of food or about its placement on the plate are not included here. Refusals and complaints can be spontaneous or in response to an offer made by the parent. Refusals to parental offers of dessert are included if the offer qualifies as a Coax. Refusals and complaints are coded even if the child eventually complies.

Examples (Verbal): You eat it. Take it away. I don't like that. I don't want anymore. But Mom! Oh Mom! in response to a Command or other prompt to eat by the parent. I'm only going to eat half my hamburger. I'm done (if food is present) No (in response to food or offer of food) Yukk (when in response to food) This tastes bad.

Examples (Non-verbal): Turning head away
Refusing to open mouth for food
Gesturing for food to be removed

REQUESTS FOR FOOD (Q):

Definition: Any verbal or nonverbal behavior initiated by the child in which the child asks for additional food or a new food without prompting from the parent. Affirmative responses to parental offers of food are not scored as requests.

Examples: For older children (**3 & above**), includes asking for or pointing at food. For younger children (**2 & below**), includes direct verbalization for food, as well as crying for or reaching for food. Crying is scored as a request when the behavior stops upon presentation of food.

Non-example: Parent asks if child wants salad or bread, and child responds "bread".

CRY (Cr):

Definition: Vocal distress, with or without tears, that is not for food. Although crying may be continuous, it is coded only once per interval. Crying that continues from one interval into the next, should be scored in both. To be coded as a cry, the vocalization must be clearly distinguished from the usual speaking tone of the child.

CHILD TALK (CT):

Definition: Any verbalization by the target child. A verbalization is defined as a complete phrase or a single word that stands alone.

Non-Verbal Child - (less than 3) talk for a non-verbal child includes any vocalization, including nonsense syllables, with the exception of wheezing or involuntary sighs while bottle feeding.

Verbal Child - (3 or older) - talk for a verbal child includes recognizable conversation Grunts are not

Recorded for verbal children.

Examples: Why do I have to eat that?
humming or singing
Nyah, nyah, nyah

Nonsense syllables by a nonverbal child

Non-examples: Nodding or other nonverbalizations (unless in the case of a nonverbal child verbalizing with "uh, uh, uh." The verbalization is coded but not the nodding, etc.)

Grunts by a verbal child

The sighing sound emitted by a child while bottle-feeding

PLAY (P):

Definition: Includes play with toys or use of food-related materials as play objects. This code is scored on an occurrence/nonoccurrence basis within an interval.

Play with Toys - includes any time a child touches, picks up, or handles an object that is clearly identified as a toy.

Examples: Child picks up a ball and throws it across the room.
Child gives bottle to a doll.

Play with Food or Related Items - includes play with food, utensils, plate, or other objects related to eating. Handling these objects is considered play when the child uses them for a purpose for which they are clearly not intended. Use of such objects must also be clearly repetitious and exaggerated to be coded.

Examples: Child makes repeated sweeping arm motion with fork in hand.
Twirling a bunch of grapes.
Repeated tossing of food up and down.
Playing "drums" on the table with a fork.
Stacking food.

Qualification: Play is coded only if the child is within reach of his/her food. Consequently, a Play and an Away cannot be coded simultaneously.

AWAY (A):

Definition: Any time the child is more than an arm's length from his/her place of food (regardless if standing or sitting), puts head below the table (defined as the mouth going below the horizontal plane of the table) or turns back to the food (defined as turning back to and being out of reach of the main source of food). Do not code an Away (A) if the plate has been removed from the table by the parent for a second helping and the child remains seated at the table.

QUALIFICATIONS:

1. A Play (P) cannot be coded while the child is Away (A).
2. If the child is off camera, parent behaviors can be coded if they are audible or visible, e.g., a command to come back to the table and eat.
3. Bites and Sips are the only child behaviors that can be coded if child is away and is visible on camera.

Examples: The child is standing on his/her chair more than arms length away from food.
The child gets up and walks away from the table.

Non-examples: The child is standing on his/her chair but is bending over and is still within reach of food.

The child stands more than an arm's length from his/her plate but takes his/her main source of food and eats while standing next to the table.

The child is seated in highchair facing forward and turns their upper body to look at something happening behind them.

The child is sitting sideways in the chair within reach his/her food.

TV WATCHING (TV):

Definition: TV is on and the child's head is oriented in the direction of the TV and he/she is staring/looking at the TV. Scored on an occurrence/non-occurrence basis within an interval.

Appendix C

Tape ID: _____ Coder Name: _____ Date: _____

Adherence Checklist: Active treatment elements

Behavior	Present	~ time
Differential Attention		
Rationale for praising		
Instruction on praising		
Rationale for ignoring		
Instruction on ignoring		
Rules and Consequences: The Basics		
Rationale for rules & consequences		
Instruction on rules & consequences		
Rules and Consequences: Privileges		
Rationale for privileges		
Instruction on privileges		
Nutrition		
Snacks		
Three per day		
Grazing undermines meal goals		
General food or strategy recommendations		
Breakfast: General food or strategy recommendations		
Lunch: General food or strategy recommendations		
Dinner: General food or strategy recommendations		

Instructions

Check the “present” column for any elements present in the session. Record the approximate minute at which the element first occurs. Subsequent time markers are not necessary.

CODING MANUAL

Behaviors

Differential Attention

Praising

Rationale

- Adult attention is highly rewarding to children
- Giving children attention when they do something you want them to do is a good way to teach them the right behaviors.

Instruction

- Instructor provides a specific description of the behavior parents are pleased to see
- Instructor provides a positive evaluation of a behavior
- Instructor provides examples of praise and non-praise
- Instructor discusses importance of immediate timing

Ignoring

Rationale

- Adult attention is highly rewarding
- Ignoring is a good way to decrease behavior
- Discussing extinction burst

Instruction

- Instructor identifies behaviors to ignore (e.g., complaints about food)
- Instructor discusses importance of watchful ignoring
- Instructor discusses importance of ignoring in conjunction with praise

Rules and Consequences: The Basics

Rules and Consequences

Rationale

- Praising and ignoring are necessary but not sufficient
- Examples of times to use rules and consequences (e.g., hitting)

Instruction

- Instructor defines a rule –the relationship between a behavior and consequence in advance
- Instructor defines a consequence –the result that will consistently follow the behavior (reinforcement/punishment)
- Instructor discusses how to implement rules (e.g., explained in advance, kept simple and clear)
- Instructor states that a meal duration rule should be instituted (i.e., after set time, meal will be taken away)
- Instructor asks parents to practice telling children the rule

Rules and Consequences: Privileges

Privileges

Rationale

- Use of privileges gives parents more leverage
- Can be used with all children in all situations
- Parents give away many rewards for free, these rewards can be identified and leveraged to reward identified behavior

Instruction

- Instructor identifies an example of a privilege
- Instructor provides an example of the way in which a privilege might be used to reinforce a behavior
- Instructor discusses ways to identify privileges (e.g., something the child will work hard to obtain or avoid losing)

Nutrition

Note: In order to identify that a meal was discussed in a session, the instructor must provide specific instruction regarding how to increase calories at that meal. The mention of the meal in passing (i.e., “that’s a great topic to discuss when we get to breakfast,” or “we will talk more about that when we get to dinner”) is not enough.

Snack

Three Per Day

- Instructor states that goal will be for parent to offer child three snacks per day

Grazing undermines meal goals

- Instructor discusses way in which grazing undermines intake goals
 - (e.g., prevents experience of hunger, fewer calories consumed overall, foods kids graze on tend not to be calorie dense)

General Food Strategy Recommendations

- Instructor discusses meal-specific recommendations for increasing calories

Lunch

General Food Strategy Recommendations

- Instructor discusses meal-specific recommendations for increasing calories

Breakfast

General Food Strategy Recommendations

- Instructor discusses meal-specific recommendations for increasing calories

Dinner

General Food Strategy Recommendations

- Instructor discusses meal-specific recommendations for increasing calories

Vita

Shannon Estelle Hourigan was born on March 29th, 1980 in Warwick, Rhode Island and is an American citizen. She graduated from Attleboro High School, Attleboro, Massachusetts in 1998. She received her Bachelor of Arts in Psychology from Vassar College, Poughkeepsie, New York in 2002 where she graduated with High Honors from the Department of Psychology and Vassar College. Between June 2003 and May 2005, she was a Study Coordinator in the Psychiatry Department at Boston University in Boston, Massachusetts, where she coordinated a variety of government and industry funded clinical trials of pharmacological treatment for substance use disorders. From May 2005 through July 2006, she worked as a Program Evaluator at the Center for Anxiety and Related Disorders at Boston University where she coordinated a treatment development study designed to improve the standard of care for adolescents with co-occurring traumatic stress and substance use. She received a Master of Science from Virginia Commonwealth University in May 2009. While at VCU, she was the recipient of the Deborah Braffman Schroeder Award for Research (2011), and she received the APA Division 54 Marion and Donald Routh Student Research Grant Honorary Mention (2010) for her work on this project. She completed her pre-doctoral internship in Pediatric and Clinical Child Psychology at the University of North Carolina-Chapel Hill in 2012.

ⁱ Two violations to fidelity were identified; however, upon review, neither instance represent a true violation. For example, in the first, privileges were identified as having been discussed and taught in session three (i.e., one session prior to their scheduled introduction). However, that session involved a broader discussion of reward and reinforcement principles and the way in which they differ from bribes following a question from a parent and did *not* involve discussion or instruction on how to implement privileges in the context of eating or meal times. In the second instance, dinner was identified as having been discussed in session four, two sessions before the dinner-focused session six. However, upon review, this conversation involved a general discussion of toddler eating habits and preferences and did not involve direct instruction for increasing calories during the dinner meal.