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INCREASING CALORIE CONSUMPTION IN CHILDREN WITH CYSTIC FIBROSIS: REPLICATION WITH 2-YEAR FOLLOW-UP

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Three mildly malnourished children with cystic fibrosis and their parents participated in a behavioral group-treatment program that focused on promoting and maintaining increased calorie consumption. Treatment included nutritional education, gradually increasing calorie goals, contingency management, and relaxation training, and was evaluated in a multiple baseline design across four meals. Children's calorie intake increased across meals, and total calorie intake was 32% to 60% above baseline at posttreatment. Increased calorie consumption was maintained at the 96-week follow-up (2 years posttreatment). The children's growth rates in weight and height were greater during the 2 years following treatment than the year prior to treatment. Increases in pace of eating and calories consumed per minute were also observed 1 year posttreatment. These findings replicated and extended earlier research supporting the efficacy of behavioral intervention in the treatment of malnutrition in children with cystic fibrosis.

DESCRIPTORS: cystic fibrosis, malnutrition, group treatment, maintenance

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Cystic fibrosis (CF) is a genetically transmitted disease that affects 1 in 2,500 live Caucasian births (Bowman & Mangos, 1976). The disease is a generalized dysfunction of the exocrine system affecting the gastrointestinal, pancreatic, hepatic, respiratory, and reproductive systems (Matthews & Drotar, 1984). It is most commonly characterized by pulmonary disease (chronic lung infections) and pancreatic insufficiency that results in inadequate digestion and malabsorption of nutrients, especially fats. With current treatment, the mean life expectancy is 29 years (CF Foundation, 1992). Chronic pulmonary disease is responsible for 90% to 95% of the deaths in CF (Wood, Boat, & Doershuk, 1976). However, nutritional status has been found to be highly correlated with pulmonary functioning and to affect recovery from acute infections (Gurwitz, Corey, Francis, Crozier, & Levison, 1979; Kraemer, Rudeberg, Hadorn, & Rossi, 1978). Despite the importance of adequate nutrition and the recent advances in treatment of CF, poor growth and malnutrition continue to be a significant problem. The CF Patient Registry reported that 40% of CF patients were below the 5th percentile weight for age in 1991 and that this rate has been relatively stable over the preceding 2 years (CF Foundation, 1992).

Inadequate weight gain in CF is believed to be the result of an energy imbalance (Durie & Pencharz, 1989). Because of the combined effects of the respiratory disease, a possible elevated metabolic rate (O'Rawe et al., 1992), and malabsorption, it has been estimated that a child with CF needs 120% to 150% of the recommended dietary allowance (RDA) of calories for healthy children (Dodge, 1988) with a normal to high (40%) fat intake (MacDonald, Holden, & Harris, 1991). Studies of severely malnourished patients have

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demonstrated that increasing their calorie intake via artificial diets (i.e., high-calorie formulas delivered orally; Allan, Mason, & Moss, 1973), parenteral supplementation (i.e., formula delivered through a vein; Lester, Rothberg, Dawson, Lopez, & Corpuz, 1986), and enteral supplementation (i.e., formula delivered by gastrostomy and nasogastrostomy tubes; Levy, Durie, Pencharz, & Corey, 1985) can result in improved weight gain. Thus, it appears that the energy demands of the disease may be offset by increased calorie intake.

However, studies assessing the oral intake of children with CF have found that they are consuming only 100% of the RDA of calories (Buchdahl, Fullylove, Marchant, Warner, & Brueton, 1989; Daniels, Davidson, & Cooper, 1987; Stark et al., 1991; Tomezsko, Stallings, & Scanlin, 1992) and, thus, are not meeting the CF calorie recommendations. The recent Consensus Conference report on nutrition and CF (Ramsey, Farrell, & Pencharz, 1992) advocated prevention and early intervention via nutritional education and behavioral intervention to promote optimal growth in all CF patients. Although nutritional education has been found to be important (MacDonald, Kelleher, & Littlewood, 1988), it has not been effective in increasing children's intake to 120% of RDA (Bell, Durie, & Forstner, 1984; MacDonald et al., 1988). Nutritional counseling alone may not be sufficient to promote optimal calorie intake because it addresses the issue of what to feed children with CF but does not address the problem of how to get these children to consume excess calories.

Possible behavioral components of inadequate calorie intake in CF were first noted by Stark, Bowen, Tyc, Evans, and Passero (1990). In an intervention study to increase calorie intake, parents of children with CF complained that their children engaged in behaviors incompatible with eating, making increased intake difficult. Specifically, they reported that their children ate slowly, complained of fullness and abdominal pain early in the meal, and that mealtimes were a "battleground." In a subsequent observational study, Stark et al. (1991) found that children with CF took twice as long to eat dinner as healthy peers did and that there was a negative correlation between eating and talking in the families of CF children, but no correlation between these behaviors for families of healthy peers was found. Recently other investigators have reported similar findings from surveys and interviews with parents of CF children. Crist, McDonnell, Beck, Gillespie, and Mathews (1992) found that parents of children with CF reported problems of excessively long meals, delay of eating by talking, and spitting out food significantly more than parents of healthy peers did. They also reported a significant negative correlation between the number of problems and calorie intake for the CF patients. Quittner, DiGirolamo, and Winslow (1991) reported that 94% of their sample of 32 parents of preschool children with CF cited mealtimes as problematic, with "child having poor appetite," "difficulty achieving a balanced diet," and "child resisting enzymes" as specific examples.

Thus, behavioral factors appear to contribute to the problem of insufficient intake in children with CF (Bowen & Stark, 1991). However, research addressing treatment of behavioral components of intake in this population is limited. Only two studies have been reported to date. Singer, Nofer, Benson-Szekely, and Brooks (1991) reported inpatient treatment of 4 severely malnourished CF patients aged 10 to 40 months with positive reinforcement of food acceptance, extinction of "negative" behaviors, and parent training. The children increased their intake from a mean of 54% to 92% of RDA, with 3 of the 4 increasing to 100% of RDA. At follow-up, 3 of the 4 children also increased their percentile of weight for height. Stark et al. (1990) investigated a group behavioral intervention for increasing calorie consumption in 5 mildly malnourished CF patients aged 6 to 12 years. These patients and their families were seen simultaneously, but in separate groups, for six outpatient group sessions. The treatment was evaluated with a multiple baseline design in which meals (snack, breakfast, lunch, and dinner) were targeted sequentially with an intervention consisting of nutritional education and behavioral management. After treatment, the children's calorie intake increased 25% to 43% and was above 120% of the RDA. The

children also showed increases in weight. Of equal importance was the maintenance of treatment gains in calorie intake at follow-up, 9 months posttreatment.

The present study was designed as a systematic replication of Stark et al. (1990), with the following changes. First, due to the lower increase in dinner calories compared to snack calories in Stark et al. (1990), the order of the meals targeted was reversed and snack was targeted last. This allowed collection of additional data on the effects of treatment versus order of intervention. Second, a session on relaxation training was added in the middle of intervention (after dinner and lunch and before breakfast and snack) to address the children's complaints of abdominal pain in association with meals. Relaxation was chosen because it has been successfully used with children experiencing abdominal pain (Sanders et al., 1989) and is hypothesized to give patients and families a sense of control over the situation as well as to reduce pain (McGrath et al., 1985). Third, the present study followed the children for a longer period posttreatment (2 years) and included a video assessment of changes in eating behaviors 1 year after treatment.

METHOD

Subjects

Three children with CF and their parents participated in treatment: Sam (3 years 11 months), Sarah (8 years 5 months), and Tess (6 years 5 months). Informed consent was obtained from each family. Two subjects were recruited from Rhode Island Hospital (RIH) CF Clinic, and 1 subject (Tess) was recruited from Massachusetts General Hospital (MGH) CF Clinic. The subjects had all participated in a study assessing eating in children with CF and healthy controls (Stark et al., 1991) and were interested in treatment. By parent report, all children exhibited behavior problems at meals. Tess's parents reported that she took an excessively long time to eat meals (i.e., approximately 60 min for dinner). Sam's and Sarah's parents reported that Sam and Sarah dawdled during their meals and terminated them early, and thus rarely ate a "sufficient" amount of food. Furthermore, Sarah's mother reported that Sarah, who sometimes vomited, decreased her intake for days afterward to avoid further vomiting.

All children had CF with chronic pulmonary disease and pancreatic involvement. Weight percentile for age was at the 12th, 10th, and 25th for Sarah, Tess, and Sam, respectively. Schwachman scores are a classification of disease status based on four categories of functioning: general activity, physical findings, nutritional status, and chest X rays. Each category is scored from 1 to 25, and the final score is the sum of the four categories. A total of 100 points is possible, with 86 to 100 being "excellent," 71 to 85 being "good," 56 to 70 being "mild," 41 to 55 being "moderate," and below 40 being "severe" (Schwachman & Kulczycki, 1958). Schwachman scores were available for Sam and Sarah and were 88 (excellent) and 68 (mild), respectively. All children were developmentally normal for age with no gross defects. Using the Hollingshead (1975) two-factor index of social status (education and occupation), the three families were all ranked as II, medium business or technician. Both Sarah and Sam were from single-parent families, and Tess resided with both natural parents.

Setting

The parents and the children were seen simultaneously once a week in separate groups. The parents were seen by a clinical psychologist and pediatric dietitian, with medical consultation available as needed from the RIH CF Center physician. The children were seen by a postdoctoral fellow in pediatric psychology and an undergraduate research assistant. The children's group took place in a hospital classroom equipped with a table and chairs, chalkboard, and a kitchen.

Dependent Measures

Calorie intake. The children's calorie intake was assessed using 7-day weighted diet diaries recorded by their parents. Parents were given food scales and trained to measure and record the children's food intake accurately during Session 1. In addition, diet

diaries were reviewed weekly by the dietitian, and the parents were provided with feedback on their accuracy. Diet diaries were kept daily for the 2 weeks between Sessions 1 and 2 (baseline), daily during intervention, and for 7 days during each follow-up at 4, 12, 24, 48, and 96 weeks. During the 2-week baseline and throughout treatment, parents calculated caloric values of all foods their children ate; at all follow-up sessions, parents were asked not to record calorie content to decrease experimental demands that might have led to inflated recordings. Nutritional content was analyzed pre- and posttreatment by entering the individual food diaries into the Nutripractor® 4000, a standard computerized nutrition program.

Anthropometric measures. The children's weights were obtained using a Seca Integra® 815 digital scale. Weights were taken by a trained assistant who followed a standardized protocol. The children's heights were obtained by the same assistant using the Shorr Infant/Child Height Measuring Board calibrated in centimeters and inches. Weights and heights were obtained in this standardized fashion at the beginning and end of intervention and at all follow-up visits. Weight was also measured weekly throughout treatment. Weights and heights were also obtained from the children's medical records in the CF clinic for 1 year prior to the study and the 2 years following treatment. Although the clinic weighing and measuring protocols were not as stringent as the research protocol, they were done on consistent weight and height scales, in a standardized fashion, and were used to evaluate the children clinically as they were followed at regular (3- to 4-month) intervals each year. Inclusion of these data allows evaluation of the treatment gains in height and weight in comparison to the child's growth for the year prior to treatment. These comparisons are important in evaluating the significance of gains made following treatment because of ongoing growth and development.

The percentage of body fat was estimated from skinfold measures taken with Lange skinfold calipers at four sites (triceps, biceps, subscapula, and suprailiac) using equations derived for children (Brook, 1971). Skinfold measures were taken at the beginning and end of treatment and at each follow-up period.

Pulmonary functioning. Because pulmonary functioning becomes diminished as CF progresses, measures of pulmonary functioning are often used as part of the global evaluation of the course of the disease. Measures of forced vital capacity (FVC), forced expiratory volume at 1 second (FEV_1), and forced expiratory flow during the middle half of the forced vital capacity ($FEF_{25\text{-}75\%}$) were used in this study. FVC is a measure of lung capacity, and FEV_1 and $FEF_{25\text{-}75\%}$ are sensitive to airway obstruction; they are used to indicate exponential decline in pulmonary functioning in large groups of CF patients (Corey, Levison, & Crozier, 1976). Pulmonary function tests (PFTs) are routinely administered on a yearly basis and whenever clinically indicated by chest infections. In the current study, PFTs were conducted by the RIH Pulmonary Department using standard spirometric techniques pre- and posttreatment and at the follow-ups conducted 48 and 96 weeks posttreatment.

Observed mealtime behavior. One videotaped dinner meal from each family prior to intervention and 1 year postintervention was scored for bites in 10-s intervals to obtain a distribution of bites throughout the meal as a measure of pace of eating. Bites were defined as taking solid food into the mouth. The food must pass the child's lips before it is considered a bite. Reliability was assessed on 100% of the videotapes by having two observers score them independently, with the primary observer being blind to the condition pre- or posttreatment. Kappas were calculated on the exact agreement on the number of bites within each interval. The average kappa across all videotapes was .88 (range, .78 to .93). Calories consumed per minute were also calculated by dividing the number of calories consumed (as recorded on the diet diary) by the number of minutes the child spent at the meal (as recorded by the videotape scoring).

One year was chosen for the posttreatment assessment of eating behavior because it was considered to be the most sensitive point at which to detect differences in mealtime behavior. Parents in

the original study (Stark et al., 1990) had anecdotally reported that 1 year following treatment their children's eating was the most "normalized." That is, their children ate independently without the need for either the prompts that parents gave prior to treatment or the behavioral treatment techniques they learned during intervention. The pretreatment videotape was randomly chosen from three tapings conducted as part of the families' participation in an assessment study on children's eating (Stark et al., 1991) before treatment. Because results from this study indicated that there were no differences in the pace of eating across three meals, only one videotaping was completed posttreatment. All meals were assessed for representativeness of the children's usual dinner behavior with parent ratings. All videotaped dinners used in the present study had been rated as "3, very similar" or "4, completely similar" on a scale of "1, not at all similar" to "4, completely similar."

Procedure

Baseline. Session 1 served as baseline for the present study. Data were obtained on all dependent measures, and diet monitoring procedures were taught to the parents. Baseline data for calorie intake consisted of food diaries kept for 2 weeks prior to the second group meeting.

Intervention. Intervention occurred during Sessions 2 through 7. The overall calorie goal was set so that each child increased his or her total calorie consumption 25% to 50% by the end of treatment. These goals are consistent with the goals used in calorie-supplementation research (e.g., Lester et al., 1986; Levy et al., 1985; Shepherd et al., 1986; Shepherd, Thomas, Bennett, Cooksley, & Ward, 1983). The overall calorie goal for each child was divided by the number of meals targeted (i.e., four), yielding an average increase of 100 to 200 calories per meal. Individualized goals were set so the children increased calories sequentially across meals and maintained their calorie intake in nontargeted meals each week.

The parent treatment group received nutritional education regarding the meal being targeted and child behavior-management strategies focusing on how to get the children to eat the foods presented. Nutritional education consisted of providing the parents with information regarding their child's calorie intake at the targeted meal as obtained from their diet diaries, the child's new calorie goal at the targeted meal, and specific recommendations on how to achieve the new goal. Recommendations included using alternate cooking methods to boost the calories of foods currently eaten by the child (e.g., frying instead of baking), adding calorie boosters (e.g., extra butter, dry milk, syrups), suggesting alternate higher calorie foods to those typically served, and providing high-calorie recipes for the targeted meal. During four of five intervention weeks, a new meal and a particular food group were targeted. For example, Session 2 targeted dinner and proteins, Session 3 targeted lunch and fruits and vegetables, Session 5 targeted breakfast and dairy products, and Session 6 targeted snacks and grains. The last session focused on maintaining the children's calorie intake on "sick days" (via high-calorie drinks) and a review of the nutritional program taught.

Child behavior-management strategies were also taught weekly. The sequence of the strategies taught followed the general principles of parent training recommended by Forehand and McMahon (1981). Specifically, parents were initially taught to use differential attention (praising and ignoring) and were gradually introduced to the use of contingent privileges (loss of a privilege contingent on not meeting a meal goal). Time-out was not taught because of its potential to function as a negative reinforcer during meals the child might prefer to avoid. As the parents learned behavior-management skills, these same techniques were simultaneously being used in the children's group. The specific sequence and content of the behavioral management techniques taught in each treatment group are described in Stark et al. (1990).

The children's treatment group received nutritional education, behavioral practice of meeting weekly calorie goals via consumption of a high-calorie meal, and a behavioral reward program that involved exchanging stars earned on a star chart for meeting weekly calorie goals for a trophy. In

addition, the children's group leaders used the same behavior-management strategies taught in the parents' group to promote appropriate behavior in the group setting and to manage the in-session meals. As with the parents' group, each children's group focused on increasing calories at a particular meal. The children were provided information on making higher calorie food choices and boosting the calories of foods they were eating at each targeted meal. Star charts were used to reinforce gradually increased eating across meals. Each week the star chart was redeemable for a trophy if the child earned 75% of possible stars. Between Sessions 1 and 2 (baseline), the children could earn stars for helping their parents record their diets. Gradually, over the course of treatment the criteria for stars increased such that after Session 2 the children had to increase their calories at dinner while keeping their calorie intake for other meals the same as they were at baseline. At Session 3, they had to increase their calories at lunch, in addition to dinner, and to continue eating the same amount as in baseline for breakfast and snack. They were also instructed on eating techniques that would increase their pace of eating and were incompatible with dawdling, such as taking two or more bites in a row, loading the fork immediately after taking a bite, and always taking a bite (and swallowing it) before talking. Sessions 5 and 6 proceeded in a similar format and targeted breakfast and snack. Session 7 focused on reviewing the children's progress in treatment, reminding them of what they needed to do to continue to achieve their calorie goals, and instructing them on how to manage "sick days." The content of the children's group, except for the addition of a session on relaxation, is also described in Stark et al. (1990).

The children were taught a relaxation exercise during Session 4 to address complaints of abdominal pain after meals. The relaxation protocol was designed for children (Koeppen, 1974); it had them imagine they were different animals as they tensed and relaxed eight muscle groups. For example, the children were asked to imagine that they were turtles, pulling into and out of their shells, when they tensed and relaxed their shoulders. An audiotape of the relaxation exercise was given to each child so he or she could practice at home. A special sticker chart was used to promote practice of the relaxation techniques during the following week in addition to continued use of the meal star charts for calorie intake.

Follow-up sessions (4, 12, 24, 48, and 96 weeks). The parents completed 7-day food diaries during the week prior to each follow-up session. Follow-up meetings were viewed as booster sessions, especially during the first 6 months. Parents were asked about the representativeness of the food diaries and their satisfaction with the program, and were helped in solving difficulties that had arisen since the last meeting. For example, at both the 4- and 12-week follow-ups, the children were exhibiting difficulties meeting calorie goals at lunch and snack following their return to school. Strategies to address the decreased lunch calories were discussed; these included the provision of high-calorie lunches by parents and the provision of consequences for the children eating them. The children's group focused on rewarding calorie and weight maintenance or gain, engaging in fun activities, and eating a high-calorie snack.

Experimental Design

The primary dependent measure was calorie intake, both across meals and total calorie intake, pre- and posttreatment. The effect of intervention on calorie intake was evaluated in a multiple baseline design across dinner, lunch, breakfast, and snack. The intervention included two primary components directly relevant to each meal—nutrition education and contingency management strategies (cf. Stark et al., 1990). In the children's group, the children were given a practice meal, and the group leaders used differential attention to reinforce increased calorie consumption. In addition, each week the children received trophies contingent upon meeting specified calorie goals. In the parents' group, the parents were given nutritional information about the targeted meal and were taught a contingency management strategy for managing their child's eating.

To evaluate the effect of treatment on the children's total calorie intake, their average daily calories pre- and posttreatment were compared via a

paired t test. To analyze further the calorie gains in the context of the CF dietary recommendations, data were also collected on the percentage of the RDA of energy each child achieved pre- and posttreatment and at the 48- and 96-week follow-ups. Nutrient analysis of each child's diet was also done pre- and posttreatment.

The physiological dependent measures (weight, body fat, height, and PFT scores) were examined to evaluate the effect of increased calorie intake on relevant health measures. Weight gain, weight percentile for age, and percentage of body fat were reported pre- and posttreatment and at the 4-, 24-, 48-, and 96-week follow-ups. In addition, weight and height were evaluated using data from each child's medical record for 1 year pretreatment and 2 years posttreatment and were compared to expected norms per age. The dependent measures of pace of eating and calories consumed per minute at dinner from the videotape assessments were examined graphically.

RESULTS

Calorie Intake

Daily calorie intake averaged across the 3 children for each of the four meals is shown in Figure 1. Average calorie intake at each meal was stable during baseline and increased only following introduction of the intervention for that meal. At each meal the children achieved or exceeded their calorie goals of 100 to 200 calories per day increase over baseline. Furthermore, the increased calorie gains were maintained at all follow-up assessments for dinner and breakfast. The decrease in lunch and snack calories at Weeks 4 and 12 of follow-up corresponded to the return to school and the resulting reduction in parental control of eating. Following a booster session on behavioral strategies during follow-up at Week 12, calorie intake increased to treatment levels at Week 24 and was maintained at Weeks 48 and 96.

Nutrient Analysis

The average calorie intake per day and the percentage of the RDA for energy these calories represent are presented in Table 1 for each child at baseline, end of treatment, and the 48- and 96-week follow-ups, along with the nutrient analysis of their diet diaries pre- and posttreatment. Posttreatment calorie intake increased by 60%, 32.6%, and 53.7% for Sarah, Tess, and Sam, respectively, and all children maintained their posttreatment calorie gains at the 48- and 96-week follow-ups. A paired t test of calorie consumption showed a significant increase in daily calorie consumption, $t(2) = 4.67$, $p < .05$.

The clinical significance of the increased calorie intake is reflected by the change in the percentage of the RDA for energy intake per child (Table 1). The RDA for energy is based on the calories per kilogram required per age level for growth and development. None of the children were consuming the CF dietary recommendations of 120% to 150% RDA of energy before treatment (Table 1). Following treatment, all of the children increased their percentage of RDA, and Sarah and Sam were above the CF recommendations. At the 48- and 96-week follow-ups, all of the children maintained their calorie gains and were within the CF recommendations. Dietary analysis also demonstrated that all the children increased their fat intake during treatment and were at the CF recommendations of a moderate to high fat intake posttreatment.

Physiological Measures

The effects of the increased calorie intake on the physiological measures of weight, weight percentile for age, percentage body fat, and pulmonary function are presented for all children in Table 2. The children gained an average of 0.66 kg during treatment. Sarah and Tess gained 1 kg and 1.2 kg, respectively, and Sam lost 0.2 kg. After medical consultation at the end of treatment, Sam's pancreatic replacement enzymes were changed to a stronger formula, and at the 4-week follow-up his weight had increased by 0.6 kg. All of the children continued to gain weight at the follow-up sessions. The increased weight resulted in improved weight percentiles for age during treatment and through the 48-week follow-up. At the 96-week follow-up, Sarah's weight percentile had declined to pretreatment levels, whereas Tess's and Sam's remained relatively stable. The children also showed

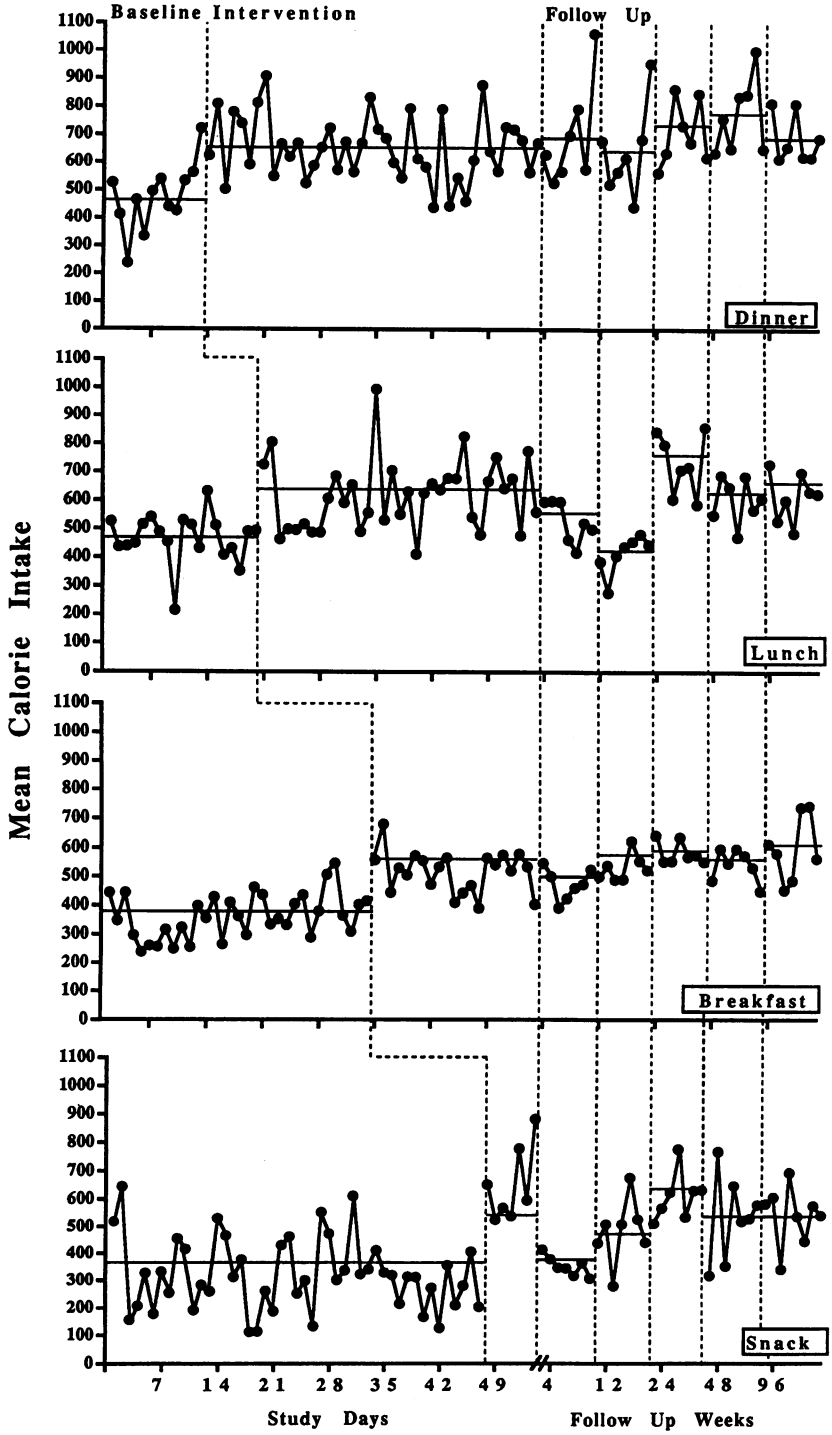

Figure 1. Average daily calorie intake across all subjects for dinner, lunch, breakfast, and snack during baseline, intervention, and follow-up weeks.

Table 1
Dietary Information for Each Subject

Subject	Measure	Baseline	End of treatment	Follow-up 48 weeks	Follow-up 96 weeks
Sarah	Average calories per day	1,747	2,799	2,415	2,774
	% RDA for energy	114	175	139	146
	% protein	14	12.1		
	% carbohydrate	55.1	52.4		
	% fat	31.8	37.2		
Tess	Average calories per day	1,453	1,921	2,212	2,206
	% RDA for energy	93.8	116	149	135
	% protein	14.8	14.5		
	% carbohydrate	48.3	44.1		
	% fat	38.6	43		
Sam	Average calories per day	1,552	2,386	2,690	2,375
	% RDA for energy	98.7	151.8	167	137
	% protein	12.5	14		
	% carbohydrate	54.8	46		
	% fat	33.5	40.8		

increases in body fat during treatment that remained above baseline levels at all follow-ups, although there was a slight decline between the 48- and 96-week follow-ups.

The results of the PFT measures are also presented in Table 2 for Sarah and Tess (Sam was not old enough for this measure). In general, Sarah and Tess showed improvement in their FVC. Tess also showed improvement in FEV_1 and $FEF_{25\text{-}75\%}$ during treatment. Sarah showed a decline on both of these measures. These measures showed some fluctuation at the 48- and 96-week follow-ups but were similar to posttreatment scores at the 96-week follow-up. The changes were not clinically significant and represent relative stability in lung function over the course of the study.

Because children are expected to grow over the course of a year, the weight gains of the children during the 1st year of the study were compared to the expected weight gains for age and sex (Fomon, Haschke, Ziegler, & Nelson, 1982) during the year before treatment, 1 year posttreatment, and 2 years posttreatment. These results are presented in Table 3. Their actual weight gain was well below the expected weight gain the year before treatment, but was above the expected gain at 1 year posttreatment for Tess and Sam. Although Sarah did not meet the expected weight gain for her age and sex during the 1st year of the study, she did show a substantial improvement over her weight gain in the previous year. During the 2nd year posttreatment, Sam continued to gain at the expected rate and Tess gained slightly less than expected. Sarah maintained her weight gain from the 1st year but did not meet the expected weight gain for her age and sex. The children's height data are presented in centimeters per year of expected versus actual height growth for the same time periods in Table 4. All of the children were below the expected growth rate for height prior to treatment and were above expected at 1 year posttreatment. Only Tess maintained this rate of growth for 2 years following treatment.

Schwachman scores for Sarah and Sam showed improvement during treatment and maintenance at the 96-week follow-up. Sarah's score increased from 68 to 82 during treatment and was 84 at the 96-week follow-up. Sam's score increased from 88 to 98 during treatment and was 96 at the 96-week follow-up.

Pace of Eating

The results of the videotape analysis of the changes in the children's eating can be seen in Figures 2 and 3. Figure 2 shows the distributions

Table 2
Physiological Measures for Each Subject

	Baseline	End of treatment	Follow-up (weeks) 4	24	48	96
Weight (kg)						
Sarah	21.8	22.8	22.8	24.0	25.0	27.1
Tess	17.2	18.4	19.0	20.0	21.2	23.4
Sam	15.4	15.2	15.8	16.2	17.8	19.2
Weight percentile for age						
Sarah	12	15	15	17	15	10
Tess	10	24	26	26	25	25
Sam	25	25	26	26	26	26
Percentage body fat						
Sarah	1.1	8.1	8.6	6.5	9.9	7.62
Tess	10.4	13	12.4	11.2	16.2	14.5
Sam	11.3	13.3	13.8	14	16.1	14.27
Pulmonary function measures[a]						
Sarah						
FVC	78	86			84	96
FEV_1	73	68			61	77
$FEF_{25-75\%}$	34	29			16	26
Tess						
FVC	85	93			88	83
FEV_1	101	120			96	85
$FEF_{25-75\%}$	77	100			75	90

[a] Pulmonary function is measured as percentage of predicted for forced vital capacity (FVC), forced expiratory volume in 1 s (FEV_1), and forced expiratory flow during the middle half of the FVC ($FEF_{25-75\%}$).

of bites during the pretreatment meal and at 1 year (48 weeks) posttreatment. Sarah's data show an increase in the number of bites at the beginning of the meal and approximately the same amount of time spent at the meal. Tess's data show an increase in the number of bites throughout the meal and a shorter meal duration. Sam's data show an increase in the number of bites at the beginning of the meal and a longer meal time.

Figure 3 shows the average number of calories

Table 3
Expected Weight Gain (Kilograms per Year) Versus Actual Weight Gain[a] per Year for Each Subject

	1 year pretreatment		1 year posttreatment[b]		2 years posttreatment	
Subject	Expected	Actual	Expected	Actual	Expected	Actual
Sarah	2.95	0.3	3.56	2.4	4.03	2.4
Tess	2.31	1.4	2.10	4.09	2.95	2.2
Sam	2.05	1.03	1.9	2.2	2.0	2.1

[a] Expected weight gain is based on norms reported by Fomon et al. (1982).
[b] The 1-year posttreatment includes the 7 weeks the children were enrolled in treatment.

Table 4
Expected Height Growth (Centimeters per Year) Versus Actual Height Growth[a] per Year for Each Subject

	1 year pretreatment		1 year posttreatment[b]		2 years posttreatment	
Subject	Expected	Actual	Expected	Actual	Expected	Actual
Sarah	5.8	4.3	5.8	7.3	6.1	4.8
Tess	6.2	6.05	6.0	6.7	5.8	6.9
Sam	8.1	7.5	7.0	7.8	6.2	4.8

[a] Expected height growth is based on norms reported by Fomon et al. (1982).
[b] The 1-year posttreatment includes the 7 weeks the children were enrolled in treatment.

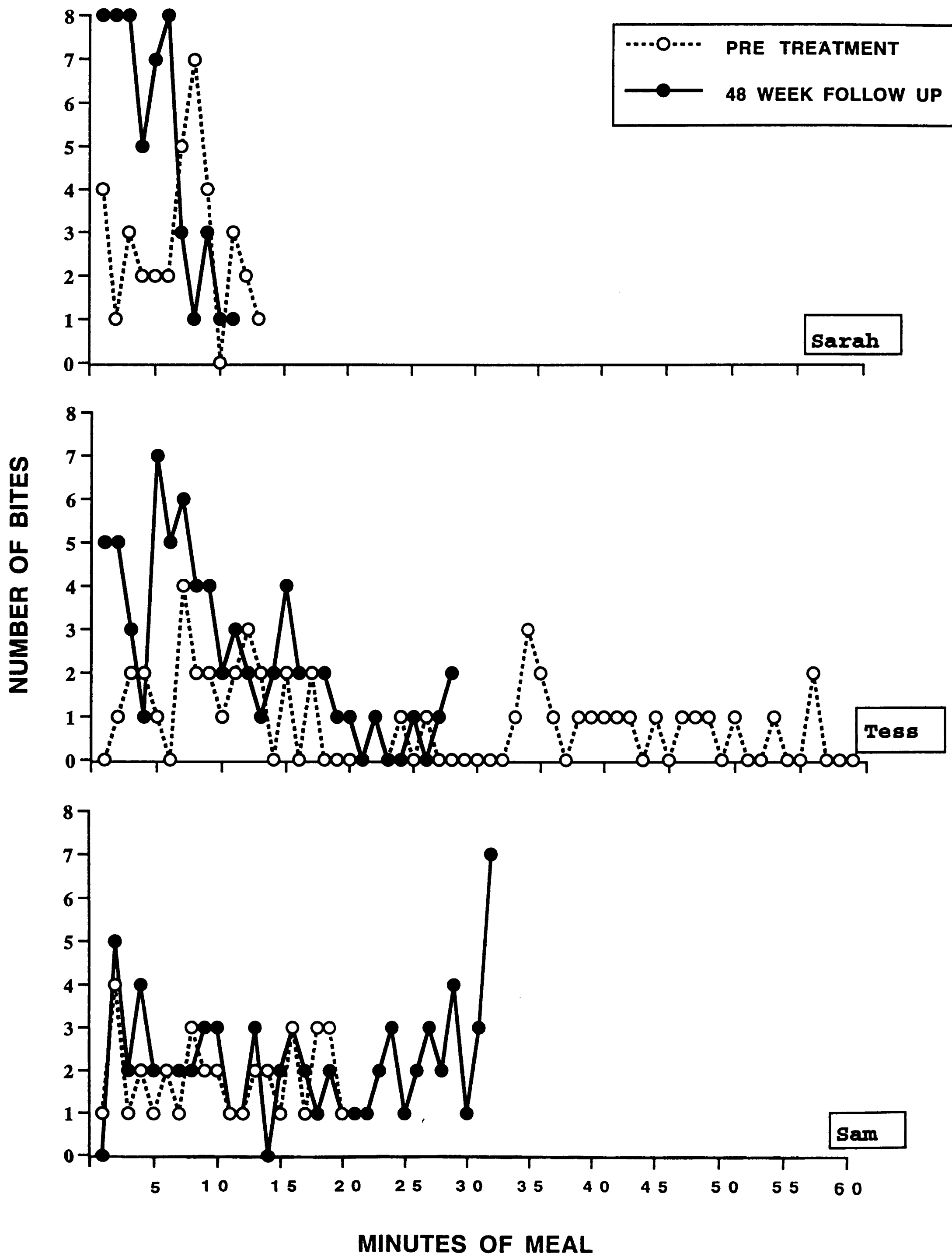

Figure 2. Pace of eating (bites per minute) during a representative dinner pretreatment and at the 48-week follow-up for Sarah, Tess, and Sam.

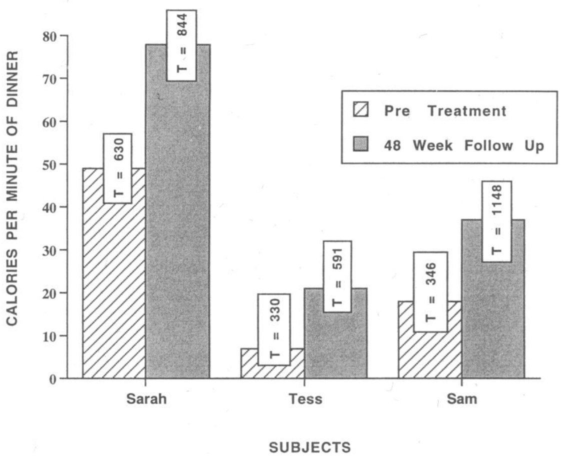

Figure 3. Calories consumed per minute and total calories (T) during a representative dinner pretreatment and at the 48-week follow-up for Sarah, Tess, and Sam.

consumed per minute at the pretreatment and 1-year (48 weeks) posttreatment meals. All of the children increased the number of calories consumed per minute 1 year posttreatment. Figures 2 and 3 show that Sarah increased her calories without increasing her meal time, Tess increased her calories and decreased her meal time, and Sam greatly increased his calorie intake (from 346 calories to 1,148 calories) while increasing his meal time by only 10 min.

DISCUSSION

The present study replicates and extends our previous research on the application of behavior analysis to calorie consumption in children with CF. As in our previous study (Stark et al., 1990), the children in the current study increased their calories an average of 46.9% across treatment sessions. The calorie increases in both studies are comparable to calorie increases produced by medical interventions (Lester et al., 1986; Levy et al., 1985; Shepherd et al., 1986). The long-term maintenance of treatment effects found in the original study were also replicated and extended in the present study. The maintenance of improved calorie intake 2 years posttreatment is important, particularly when compared to medical intervention studies in which many patients have decreased appetite during treatment (Allan et al., 1973; Levy et al., 1985) and return to baseline weight after treatment termination (Mansell et al., 1984).

The comprehensive nature of the current intervention, which included both nutrition education and child behavior-management training, precludes assessment of the relative contributions of any one component to treatment success or maintenance of treatment gains. However, previous research indicates that nutrition counseling alone either does not produce substantial calorie gains (Bell et al., 1984) or takes several years to do so (Luder, Kattan, Thorton, Koehler, & Bonforte, 1989). Although behavioral intervention alone has not been explored in this population, it is unlikely that teaching parents how to manage their child's behavior at meals without providing guidance on how to optimize the calorie content of the foods provided would result in such substantial calorie gains in underweight children with cystic fibrosis.

In addition to providing the parents with nutritional information and child behavior-management procedures, it should be noted that presentation of the nutritional information and teaching the behavioral skills in the current treatment were also conducted in a systematic manner. For example, in the parents' group, only one meal and one set of parenting behaviors were taught per session, and the information presented in each session built upon the knowledge and skills learned in previous sessions. Parental implementation of the behavioral program occurred gradually, and the contingencies used by the treatment personnel were faded over the course of intervention. Similarly, the children were given more than information on appropriate food choices. They were given exposure to these foods and opportunities to practice eating them in session each week. Initially, staff members rewarded the children for meeting their calorie goals in sessions throughout treatment; subsequently, the parents provided the rewards. Also, both groups were complementary (e.g., parent training was supported by application of the described techniques in the children's group).

The reasons for the long-term maintenance of treatment effects are unclear. We hypothesize that directly changing the parent–child interaction at mealtime may have attenuated the reinforcers associated with refusing food or dawdling at meals through the use of extinction (removal of parent attention) and punishment (loss of privileges for not meeting calorie goals). Before treatment, one of Tess's parents remained "trapped" at the dinner table trying to convince Tess to eat more of her dinner after the family had finished. This extra attention may have reinforced Tess's dawdling and slow pace of eating. When her parents began setting a time limit for meals, they were initially fearful that Tess would eat less. However, when the consequences for dawdling became termination of the meal (regardless of completion) and loss of privileges, Tess quickly increased her pace of eating and ate more food in less time. Long-term maintenance may have also been enhanced by the gradual increase in calories, leading to increases in the child's satiety threshold, which in turn allowed him or her to consume more calories before feeling full (see Bowen & Stark, 1991, for discussion).

Finally, conditioned responses to food may have changed over treatment. For example, before the intervention, Sarah's mother reported examples of conditioned taste aversions (CTAs) that Sarah developed after any episode of vomiting. Sarah's mother stated that Sarah would avoid not only the particular food associated with the vomiting but all food for several days after vomiting. During treatment, Sarah was instructed to maintain her calories after vomiting by finishing her meal or eating another meal shortly after vomiting. Stars were withheld after a meal in which she vomited unless she followed these directions. Sarah's mother reported that an important benefit of treatment was that Sarah no longer avoided food following a vomiting episode.

All of these proposed mechanisms depend upon the occurrence of change in the parent–child interaction as a result of treatment. Although the parents were taught behavior-management skills, we did not assess the extent to which parents actually implemented these skills. Measures of integrity of the independent variables should be included in future research so that functional components of treatment and its maintenance may be identified.

The pace of eating and calories consumed per minute were assessed by videotaping a typical dinner. The results indicated that children consumed more calories per bite posttreatment and appeared to have increased their pace of eating. Videotape assessment may also be an effective way of conducting measures of changes in parent–child interactions during intervention.

One important aspect of interventions in behavioral pediatrics is their effect on child health status. The most direct physiological outcomes of the present study were the anthropometric measures of weight, height, and body fat. Two of the 3 children showed increases in weight during treatment. The 3rd child showed weight gain at the 4-week follow-up after medical consultation successfully titrated replacement enzymes for digestion. All of the children continued to gain weight at the follow-up sessions, and the significance of this weight gain was shown when the children's weight gains were compared to their weight gains the year before treatment and to expected weight gain for age. The children's weight gains were most similar to those expected during the year they participated in treatment. Height growth and body fat also improved throughout the 1st year after treatment. Height growth for Sarah and Sam did not maintain the catch-up rate observed during the treatment year, and body fat was slightly lower during the 2nd year posttreatment for all of the children. However, all of these measures continued to be above baseline 2 years after treatment.

The measures of pulmonary function and Schwachman scores are less directly related to increased calories but are hypothesized to improve with nutritional status. Here the results were less clear. Although Tess showed improvement on all pulmonary measures during treatment and a slight decline across the 2 years following treatment, none of these changes were clinically significant and reflect a general stability in lung functioning. Simi-

larly, although Sarah showed improvement on one pulmonary measure and a slight decrease on two other measures during treatment, these changes were not clinically significant and reflected stability across all PFT measures during treatment and across the 2 years after treatment. Although some studies (e.g., Shepherd et al., 1986) have reported improved pulmonary functioning, the more frequently reported finding is maintenance of baseline pulmonary functioning (Mansell et al., 1984), indicating a slowing of decline in pulmonary functioning following intervention. On the Schwachman rating scale, both Sarah and Sam showed improvement during treatment and maintenance at the 2-year follow-up, reflecting an overall improvement in their physical presentation.

Clearly, the children demonstrated an overall improvement on physical measures during treatment. However, it is difficult to discern the significance of these improvements over the long term in single-subject studies with CF patients because of the progressive nature of the disease. As stated previously, CF is a complex disease with decline in pulmonary measures and weight associated with age and disease severity. As in the present study, previous studies have found that nutritional intervention is associated with a slowed rate of decline when patients serve as their own controls and their progress posttreatment is compared to their pretreatment medical records (Boland, Patrick, Stoski, & Soucy, 1987; O'Loughlin et al., 1986; Stark et al., 1990). In one of the few studies using a control group, Dalzell et al. (1992) reported similar results. In their study, CF patients receiving enteral supplementation were found to show a slower rate of decline on measures of pulmonary functioning, were less underweight, and had fewer deaths in the posttreatment follow-up when compared to a no-treatment control group. Additional research with matched controls is needed to evaluate adequately the long-term outcomes of both behavioral and medical interventions in the area of CF and nutrition, because numerous factors can affect the individual patient's long-term outcome. These factors include disease severity, activity level, increases in metabolic demand following refeeding (Vaisman, Clarke, & Pencharz, 1991), and the general progressive nature of the pulmonary disease.

In conclusion, as the survival time of CF patients increases and as more advances are being made against the lung disease (via lung transplants and genetic research), combating malnutrition through promotion of normal growth and enhanced nutrition is of increasing importance (Ramsey et al., 1992). The present study demonstrates that a combined behavioral and nutritional intervention program can be effective in increasing calorie intake. Increased calories were associated with increased body weight, height growth, body fat, and stability in pulmonary functioning. Future research should address limitations of this study by targeting a larger sample and including measures of treatment implementation and evaluation of a control group. Successful maintenance of treatment gains in calorie intake and continued weight gain 2 years posttreatment suggest that a behavioral approach to nutrition intervention holds considerable promise for addressing poor weight gain and malnutrition in children with CF.

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