

Behavioral Therapy for Children with Avoidant/Restrictive Food Intake Disorder Dependent on Tube or Oral Enteral Nutrient Formula: A Feasibility Study

KAZUMI TOMIOKA*, MASAHIRO NISHIYAMA, SHOICHI TOKUMOTO,
HIROSHI YAMAGUCHI, KANDAI NOZU and HIROAKI NAGASE

*Department of Pediatrics, Kobe University School of Medicine, Kobe, Japan; *Corresponding author*

Received 2 September 2021/ Accepted 22 December 2021

Keywords: Avoidant/restrictive food intake disorder, Behavioral therapy, Eating disorders, Tube feeding

In children with eating disorders, nutritional status and growth may depend on enteral nutrient formula. Ultimately, its goal is to introduce or reintroduce oral feeding. Japanese research on the treatment of tube or oral formula-dependent children is scarce. This study determined the feasibility of behavioral therapy for children with avoidant/restrictive food intake disorder and dependency on the tube or oral enteral nutrient formula in Japan. Medical records of children diagnosed with this disorder, dependent on the tube or oral enteral nutrient formula and who had received behavioral therapy intervention to withdraw from the formula were retrospectively investigated. We collected their characteristics at first visit and the caloric percentage from oral food intake six months after starting the treatment. In total, four patients (age range: 2–5 years) participated in this study. The feeding routes employed before the intervention were a nasogastric tube for one patient, a gastrostomy bottom for the other patient, and oral formula for the remaining patients (i.e., two children). At the sixth month of the behavioral treatment, none of the patients needed the formula, and the caloric percentage of required nutrition from oral food intake was 100%. Our data demonstrate that this behavioral therapy is feasible for children with avoidant/restrictive food intake disorder dependent on the tube or oral formula in Japan.

INTRODUCTION

Feeding problems, such as those associated with avoidant/restrictive food intake disorder (ARFID), impact nearly 5% of children and are common concerns in pediatric settings [1, 2]. According to the Diagnostic and Statistical Manual of Mental Disorders (DSM-5), clinical manifestations of ARFID include faltering growth, significant nutritional deficiencies, reliance on enteral feeding, or oral nutritional supplementation to satisfy energy needs [3]. Estimates confirm that 40–70% of children with chronic medical concerns (e.g., congenital or acquired respiratory, cardiac, and gastrointestinal problems) experience feeding difficulties [4]. These medical problems may facilitate conditioned food aversion by pairing unpleasant consequences, such as pain, nausea, or fatigue, with eating [5, 6].

For ARFID, which seriously compromises a child's nutritional status and growth, the tube or oral enteral nutrient formula is used [7]. Although the enteral nutrient formula can have significant immediate benefits [8], the ultimate goal of treatment is to introduce or reintroduce oral feeding [7].

Past studies on tube-dependent children found that behavior-based treatments are effective in weaning gastrostomy tube (GT)-dependent children in the US and Canada [7–9]. However, the effects of psychotherapy (e.g., behavioral therapy) vary by culture, and relevant research is still scarce in Japan. This research was a retrospective study comprising four cases to assess the feasibility of incorporating behavioral therapy for children with ARFID dependent on the tube or oral enteral nutrient formula in Japan.

MATERIALS AND METHODS

Patients

Inclusion criteria were patients who (1) had visited Kobe University Hospital between January and December 2020, (2) were diagnosed with ARFID by DSM-5, (3) were diagnosed with selective eating by the Great Ormond Street Criteria [4] or rely completely on tube feeding, and (4) had stable general condition through enteral nutrient formula. This study focused on four children with ARFID.

Methods

This study was a retrospective analysis of medical record data aggregated from consecutive cases. Patient characteristics were assimilated through medical records. We evaluated the caloric percentage from oral food intake, percentage of ideal body mass index (BMI), duration of inpatient hospitalization, duration of complete calorie intake through oral consumption, number of step-down treatment events, and unfavorable behaviors at mealtime or other new behavioral problems after six months of treatment.

Behavioral feeding intervention

We modified the behavioral therapy reported by Taylor *et al.* in 2019 [10]. Patients and caregivers could choose to be inpatients or outpatients, but the treatment content was the same even though only outpatient treatment was included in the original report. The doctor recommended that the caregiver be hospitalized with patients as much as possible because visits to the outpatient department occurred once every one or two months, and the treatment period was prolonged. The choice between hospitalization and outpatient care depended on familial circumstances; the children needed to be hospitalized with caregivers if they were young.

The dietary progress depended on the individual child as per the original report; however, in our behavioral treatment, the amount of food served was divided into eight dietary steps (refer to Table I). The dietary steps were suggested to patients and their parents by a developmental behavior pediatric clinician during interviews and were implemented if patients and parents agreed. That is, they were allowed not to take a step or to take one or more steps in each interview. At the beginning of the treatment, the doctor ensured that the patients were able to eat, as no problem with the swallowing and chewing function was detected. The doctor started with a minimal diet and tried to form a rapport with the patients and caregivers. Inpatients were interviewed each week, and outpatients were interviewed each month. Furthermore, the dietary steps were decreased if the prepared meals were not finished but could be raised if finished for one week in a row. The meal content started with staple food (e.g., rice, bread, and pasta), and a side dish was introduced once the staple food was properly integrated. If the children had difficulty in eating solid foods, a paste was incorporated instead. Positive reinforcement included marking days of full eating on the calendar and immediate verbal praise for desirable feeding behaviors, including bite acceptance, chewing, swallowing, self-feeding, and use of appropriate table manners. No negative reinforcement was observed.

Furthermore, the doctor provided treatment education to the caregivers. Treatment education included the following directions: first, the caregivers were not to ask patients about their diet, and they were to praise good behavior and disapprove of bad behavior. Second, the patient's mealtime, sitting position, and tableware were to be fixed as much as possible. Third, when patients were unable to complete their meals, the caregiver was to tell them that they may not be able to eat because of an illness (i.e., an eating disorder) but would be able to complete the meal by continuing treatment.

Table I. Behavioral treatment steps

Dietary step	The caloric percentage of required nutrition from oral food intake	Target meal time
Step 1	0.5–1%	<30 minutes
Step 2	1–3%	“
Step 3	3–5%	“
Step 4	5–10%	“
Step 5	10–30%	“
Step 6	30–50%	<1 hour
Step 7	50–80%	“
Step 8	100%	“

Ethics

This study was approved by the institutional review board of the hospital, and written consent from the parents and verbal assent from the children (as appropriate) were obtained.

RESULTS

Patients' characteristics

Altogether, four patients were included (three boys and one girl) in this study. Demographics and tube or oral elemental nutrition feeding data are shown in Table II. The mean age at first visit was 4 years (range: 2–5 years). The range of gestational week and birth weight was 27–42 weeks and 528–3456 g, respectively. Patients had gastroesophageal reflux (three patients), congenital heart disease (one patient), chronic lung disease (one patient),

BEHAVIORAL THERAPY AND ARFID

kidney failure (one patient), food allergy (two patients), intellectual disability (two patients), and autism spectrum disorder (three patients). The mean developmental quotient was 68 (range: 50–98), and the percentage of ideal BMI during the first visit was 75–92%. The caloric percentage of required nutrition from oral food intake was 0–10% at first visit for all patients. Patients undertook enteral nutrient formula through the oral intake (two patients), a nasogastric tube (one patient), and gastrostomy button (one patient). All children received outpatient speech therapy, including oral stimulation, and one patient received home speech therapy. Unfavorable behaviors at mealtime were recognized for all patients (e.g., vomiting, sobbing, bruxism, swallowing air, forcing others to eat, pretending to chew, and shaking their head). A patient (Case 3) had to undergo outpatient treatment due to severe food allergies and because food from outside was prohibited at the hospital.

Table II. Patients' characteristics

Case	Case 1	Case 2	Case 3	Case 4
Age at visit	2	4	4	5
Sex	Female	Male	Male	Male
Gestational age (weeks)	27	42	33	40
Body weight at birth (g)	528	3456	2040	3044
Medical history	GER, congenital heart disease, chronic lung disease	GER, food allergy	GER, kidney failure (transplantation), food allergy	None
Developmental disorder	ID	ASD, ID	ASD	ASD
Developmental Quotient	50	52	83	98
Body weight at visit (kg)	6.9	14.2	16.5	23.4
Percentage of ideal BMI at visit (%)	75	80	82	92
The caloric percentage from oral food intake	0	0	0	10% (only scone)
Age at starting with enteral nutrient formula	1 year old	2 years old	1 year old	1 year old
Route of formula intake	Oral	Nasogastric tube	Gastrostomy button	Oral
Previous intervention, frequency	Outpatient speech therapy, 2/months	Outpatient speech therapy, 2/months	Outpatient speech therapy, 2/months, home speech therapy, 1/week	Outpatient speech therapy, 2/months
Duration of previous intervention	1 year	2 years	3 years	4 years
Unfavorable behaviors at mealtime	Vomiting, sobbing, swallowing air, shaking head	Vomiting, sobbing, bruxism, swallowing air, forcing others to eat, pretending to chew	Vomiting, sobbing, swallowing air, forcing others to eat, pretending to chew	Vomiting, sobbing, forcing others to eat

GER: gastroesophageal reflex, ID; intellectual disability, ASD; autism spectrum disorder

Outcomes at six months

Two inpatients and two outpatients were enrolled in this study (refer to Table III). The caloric percentage of required nutrition from oral food intake was 100%, and the enteral nutrient formula was no longer needed by the patients after six months. The duration of hospitalization was between four and eight weeks, and the caloric percentage from oral food intake was 100% at discharge in both inpatients. The duration for achieving full-calorie

oral food intake ranged from four weeks to five months. Case 1 finished in four steps (steps 2→4→6→8) of her intervention. The percentage of ideal BMI was 66–76% at the sixth month of treatment. The unfavorable behaviors at mealtime, which existed before the intervention, disappeared in all cases, and no new behavioral problems emerged.

Table III. Outcomes at six months

Case	1	2	3	4
Inpatient/outpatient	Inpatient	Inpatient	Outpatient	Outpatient
The caloric percentage from oral food intake at 6 months	100%	100%	100%	100%
The caloric percentage from enteral nutrition at 6 months	0	0	0	0
Percentage of ideal body mass index at 6 months (%)	73	73	66	76
Body weight gain at 6 months from first visit	+0.2kg	+0.1kg	+0.5kg	-0.6kg
Height increase at 6 months from first visit	+0.8cm	+0.9cm	+4.0cm	+2.2cm
The duration of hospitalization	4 weeks	8 weeks	-	-
The duration for achieving the full calorie of required nutrition from oral food intake	4 weeks	8 weeks	5 months	3 months
Number of step-down events	None	None	None	None
Unfavorable behaviors at mealtime	None	None	None	None
New behavioral problems	None	None	None	None

DISCUSSION

In this study, the behavioral treatment enabled all four patients with ARFID to consume their diet orally and withdraw from the formula. The structural environment and targeted behavioral interventions produced outcomes that demonstrated patients' potential abilities [7].

In our study, all the patients achieved the oral nutritional goals. In previous reports of behavioral therapy for children depending tube or formula, the percentage of patients weaned from enteral feeds ranged between 43% and 100% [7–9, 11]. For example, a report with a larger sample size found that, among 77 patients dependent on tube feeding, the mean oral intake percentage increased from 30% (standard deviation [SD] 2.5%), to 82% (SD 3%) [7]. Moreover, the mean treatment duration was previously reported to be 5–46 days [5, 7, 8, 12], and inpatient treatment tended to be shorter than outpatient treatment. In our cases, the treatment duration was shorter for inpatients than outpatients. Therefore, behavior therapy in patients with ARFID dependent on the tube or oral enteral nutrient formula may be as effective in Japan as it is overseas; however, these four cases may just have happened to be successful, so further studies are needed.

Three possible reasons may explain why this study was as effective as previously reported overseas. First, we demonstrated that the caregivers were able to implement treatment procedures with high fidelity. The original and our modified behavioral interventions were found to be effective with fewer resources and lower intensity than previous ones [10]. We also considered it important that the caregiver should be prepared to provide food and proceed with the designed steps before starting treatment and that treatment consent be obtained.

Second, the reduction in behavioral problems of all participants was thought to have helped caregivers perform treatment procedures. Behavioral feeding problems frequently involve avoidance behaviors, such as food refusal and tantrums [13] secondary to a history of aversive feeding experiences (e.g., gagging, choking, painful swallowing) [14]. Additionally, the steady and consistent delivery of nutritional needs via tube feeding minimizes feelings of hunger, thus diminishing the natural motivation for oral intake [15]. As they presented unfavorable behaviors, including refusal to swallow contents orally, vomiting, bruxism, and shaking their head, as in previous reports [7], these unfavorable behaviors would have made it difficult for parents to encourage their children to eat orally. We provided treatment education to caregivers on praising the child's good behavior and disapproving of their bad behavior. As a result, the problem of extinction bursts did not appear and the behavioral feeding problems were reduced in our cases.

Third, it was possible that visually knowing the intake amount by numerical value and appearance made it easier for children, especially ASD children, and their families to set and achieve a target amount by dividing the food amount into eight steps. As previously reported, our participants had a variety of comorbid conditions, such as gastrointestinal disorders, neurodevelopmental disorders, cardiorespiratory disorders (e.g., congenital heart disease, bronchopulmonary dysplasia) and autism spectrum disorder that prevented them from establishing normal feeding [11, 7]. Visual cues may be more effective for learning/retrieval, as demonstrated by previous

BEHAVIORAL THERAPY AND ARFID

studies that used pictorial prompts for teaching children with ASD [16], so presenting the amount visually may have been effective for these children.

The children in our study were representative of those who failed to make progress when treated without receiving appropriate behavioral techniques; however, as demonstrated by the present results, even those difficult-to-treat children can make significant progress toward tube or oral formula independence when treated with an intense regimen of behavioral techniques. However, in our cases, although body weight gain did not decrease drastically, the percentage of ideal BMI at six months from the first visit had decreased. A larger trial would permit a more rigorous evaluation of weight status, suggesting that the intervention may improve growth or protect children from a possible decline in BMI-for-age percentile [17]. Therefore, we should have reduced the enteral formula more slowly.

Limitations to our study included retrospective, single facility data collection and small sample size; consequently, this topic needs to be studied prospectively in a greater number of cases and multiple facilities.

To conclude, our data demonstrated that this behavioral intervention may be feasible for Japanese children with ARFID dependent on a tube or oral formula. However, we need to increase the number of cases and conduct prospective studies in the future.

ACKNOWLEDGMENT

The authors thank the medical nursery teacher, nurses, physicians and clerks of the Kobe University Hospital who took great care of the patients and caregivers. We also thank the children and their parents for their kind cooperation.

CONFLICT OF INTEREST

The authors declare no conflicts of interest associated with this article.

REFERENCES

1. **Satter, E.S.** 1990. The feeding relationship: problems and interventions. *J Pediatr* **117**: 181-189.
2. **Lukens, C.T., and Silverman, A.H.** 2014. Systematic review of psychological interventions for pediatric feeding problems. *J Pediatr Psychol* **38**: 903-917.
3. **American Psychiatric Association.** 2013. Diagnostic and Statistical Manual of Mental Disorders, 5th ed. American Psychiatric Publishing, Washington, USA.
4. **Lask, B., and Bryant-Waugh, R.** 2013. Overview of eating disorders in childhood and adolescence. In: *Eating Disorders in Childhood and Adolescence*, 4th edition, p.33-49. Routledge, London, UK.
5. **Hyman, P.E.** 1994. Gastroesophageal reflux: one reason why baby won't eat. *J Pediatr* **125**: 103-109.
6. **Sharp, W.G., Jaquess, D.L., Morton, J.F., and Herzinger, C.V.** 2011. Pediatric feeding disorders: A quantitative synthesis of treatment outcomes. *Clin Child Fam Psychol Rev* **13**: 348-365.
7. **Silverman, A.H., Kirby, M., Clifford, L.M., Fischer, E., Berlin, K.S., Rudolph, C.D., and Noel, R.J.** 2013. Nutritional and psychosocial outcomes of gastrostomy tube-dependent children completing an intensive inpatient behavioral treatment program. *J Pediatr Gastroenterol Nutr* **57**:668-672.
8. **Byars, K.C., Burklow, K.A., Ferguson, K., O'Flaherty, T., Santoro, K., and Kaul, A.** 2003. A multicomponent behavioral program for oral aversion in children dependent on gastrostomy feedings. *J Pediatr Gastroenterol Nutr* **37**: 473-480.
9. **Benoit, D., Wang, E.E.L., and Zlotkin, S.H.** 2000. Discontinuation of enterostomy tube feeding by behavioral treatment in early childhood: A randomized controlled trial. *J Pediatr* **137**: 498-503.
10. **Taylor, S., Purdy, S.C., Jackson, B., Phillips, K., and Virues-Ortega, J.** 2019. Evaluation of a home-based behavioral treatment model for children with tube dependency. *J Pediatr Psychol* **44**: 656-668.
11. **Sharp, W.G., Volkert, V.M., Scahill, L., McCracken, C.E., and McElhanon, B.** 2017. A systematic review and meta-analysis of intensive multidisciplinary intervention for pediatric feeding disorders: How standard is the standard of care? *J Pediatr* **181**: 116-124.
12. **Greer, A.J., Gulotta, C.S., Masler, E.A., and Laud, R.B.** 2008. Caregiver stress and outcomes of children with pediatric feeding disorders treated in an intensive interdisciplinary program. *J Pediatr Psychol* **33**: 612-620.
13. **Silverman, A.H.** 2010. Interdisciplinary care for feeding problems in children. *Nutr Clin Pract* **25**: 160-165.
14. **Piazza, C.C., and Carroll-Hernandez, T.A.** 2004. Assessment and treatment of pediatric feeding disorders. In Tremblay, R.E., Barr, R.G., Peters, R.V. (ed.), *Encyclopedia on Early Childhood Development*. Centre of Excellence for Early Development, Montreal, Canada.

15. **Linscheid, T.R., Budd, K.S., and Rasnake, L.K.** 2003. Pediatric feeding problems, p. 481-198. In Roberts, M.C., (ed.), *Handbook of Pediatric Psychology*, 3rd ed. Guilford Press, New York, USA.
16. **Cohen, S., Koegel, R., Koegel, L.K., Engstrom, E., Young, K., and Quach, A.** 2021. Using Self-Management and Visual Cues to Improve Responses to Nonverbal Social Cues in Adults With Autism Spectrum Disorder. *Behav Modif* doi: 10.1177/0145445520982558
17. **Sharp, W.G., Stubbs, K.H., Adams, H., Wells, B.M., Lesack, R.S., Criado, K.K., Simon, E.L., McCracken, C.E., West, L.L. and Scahill, L.D.** 2016. Intensive, manual-based intervention for pediatric feeding disorders: Results from a randomized pilot trial. *J Pediatr Gastroenterology & Nutrition* **62**: 658-663.