A new cognitive behavior therapy for adolescents with avoidant/restrictive food intake disorder in a day treatment setting: A clinical case series

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Abstract
Objective: Avoidant/restrictive food intake disorder (ARFID) is a new diagnosis in the DSM-5 Feeding and Eating Disorders section, for which very limited treatment research has been carried out, yet. A new, 4-week exposure based cognitive behavioral therapy (CBT) day treatment, which integrated the inhibitory learning principles, was developed for adolescents with ARFID, and tested in the current study.

Method: A nonconcurrent multiple baseline design was used in a clinical case series of eleven 10- to 18-year-old patients. After baseline, the 4-week CBT followed. Measurements of DSM-5 ARFID diagnosis, food neophobia and related measures such as body weight and length, were taken at baseline (t1), at the end of the 4-week intensive day treatment (t2) and 3 months after treatment (follow-up, t3). A food selectivity test, a 1-week food diary, and behavioral measures on food intake were also taken at baseline and at 3-month follow-up. Furthermore, continuous measurements of believability of dysfunctional cognitions, anxiety, and food acceptance were taken throughout the 4-weeks day treatment.

Results: At follow-up, 10 out of 11 patients were in remission and had a healthy body weight and an average, age-adequate nutritional intake. For most patients, food neophobia scores decreased to a nonclinical range. The belief in dysfunctional cognitions and anxiety levels decreased during treatment.

Discussion: This new exposure-based CBT for adolescents with ARFID seems promising. These results may be very useful for clinical practice and stimulate further development of effective CBT interventions in the area of ARFID.

KEYWORDS
adolescents, ARFID, avoidant/restrictive food intake disorder, cognitive behavior(al) therapy, eating disorders, expectancy violation, exposure

1 | INTRODUCTION

Selective and restrictive eating in children and adolescents are increasingly recognized. Until recently, limited agreement existed on the identification and classification of these feeding and eating problems (Bryant-Waugh, Markham, Kreipe, & Walsh, 2010). The Diagnostic and Statistical Manual of Mental Disorders, 5th edition (DSM-5; APA, 2013) now includes avoidant/restrictive food intake disorder (ARFID),...
an eating or feeding disturbance where the individual fails to meet nutritional needs, leading to low weight, nutritional deficiency, dependence on supplemental feedings, and/or psychosocial impairment.

Selective and/or restrictive eating are not just problems in childhood, but can persist into or present in adolescence and adulthood. To date, only few empirical studies addressed treatment for individuals with these problems. That is, studies focused particularly on pediatric feeding disorders (e.g., Sharp et al., 2016), and many concern feeding problems before they were named ARFID; fear of eating is usually treated by systematic desensitization techniques where escape extinction, distraction, and/or relaxation techniques are used. Escape extinction, hindering the child to avoid, or escape the nutritional requirement, turned out to be very effective in decreasing anxiety-driven food refusal in young children, but its use in older children seems less appropriate. The effects of behavioral treatments for feeding problems in (young) children were reviewed (e.g., Bachmeyer, 2009; Sharp, Jaquess, Morton, & Herzinger, 2010; Williams, Field, & Seiverling, 2010). Interestingly, our specialized treatment facility has been treating severe food refusal before it was called ARFID for over 20 years and developed a stepwise behavioral therapy program for young children (De Moor, Didden, & Korzilius, 2007; Didden, Seys, & Schouwink, 1999). For adolescents, however, there are hardly any evidence-based treatment programs available.

Cognitive behavioral therapy (CBT) is an effective treatment for eating disorders (Hay et al., 2014; Murphy, Straebler, Cooper, & Fairburn, 2010; NICE, 2017). Further, because eating disorders are accompanied by anxiety (Pallister & Waller, 2008; Swinbourne & Touyz, 2007), it seems plausible to treat ARFID with similar techniques as used for anxiety disorders. Within CBT programs, exposure and behavioral exercises are powerful techniques for reducing anxiety and catastrophic thinking (Craske et al., 2008).

Craske, Treanor, Conway, Zbozinek, and Vervliet (2014) suggest that exposure therapy works through inhibitory learning, indicating that expectancy violation (and not fear decrease) should be its main objective. Under the inhibitory framework, structuring exposure exercises to maximize the discrepancy between the patient’s expected outcome and the actual outcome, the variability in the level of fear, and the number of cues targeted and contexts in which learning is practiced, will best facilitate corrective learning and decrease chances of retrieval of the original association (Craske et al., 2014). In a similar vein, Reilly, Anderson, Gorrell, Schaumberg, and Anderson (2017), suggested expanding exposure-based interventions for eating disorders (EDs) specifically. The importance of expectancy violation during exposure was already demonstrated in adolescent and adult overeating disorders (Schyns, Roefs, Mulkens, & Jansen, 2016; Schyns, Roefs, Smulders, & Jansen, 2018).

Following from this, exposure to the feared situation (i.e., eating the feared food), will be effective when the new association (e.g., “this food will not make me choke”) becomes stronger than the old association (e.g., “this food will make me choke”). During exposure, patients should detect a mismatch between their expectancies (what might happen) and the outcomes (what actually happens). To this end, it is important to detect patients’ specific expectancies and to record the belief in these expectancies before exposure starts. This technique can help to reduce catastrophic thinking (Craske et al., 2014; Schyns et al., 2016; Schyns, Roefs, et al., 2018; Schyns, van den Akker, Roefs, Hilberath & Jansen, 2018).

Based on clinical presentations, DSM-5 proposes three primary presentations of ARFID, including an apparent lack of interest in eating, sensitivity to the sensory characteristics of food, and fear of aversive consequences of eating. Despite insufficient evidence to support specific subtypes based on different etiology, hypotheses were formulated. For example, Thomas et al. (2017), designed a three-dimensional model to explain these three ARFID presentations, and speculated about implications for treatment. To design targeted interventions for ARFID, it may be needed to take these subtypes into account (Thomas, Wons, & Eddy, 2018). Thomas and Eddy’s (2019) approach is currently under investigation.

Thus, implementing the new exposure techniques to modify all ARFID-presentations seems quite challenging. Whereas some presentations of ARFID are clearly anxiety-driven (e.g., specific phobia of choking/vomiting), and appear to suit Craske’s et al. (Craske et al., 2014) theory quite well, not all presentations appear to be primarily characterized by anxiety. For patients with a lack of interest in food/feeding and whose restrictive eating is driven by disgust or aversion, this learning paradigm might seem more challenging. Although (some) ARFID presentations might be more grounded in psychological responses other than anxiety, violating the expectancies of these ARFID subtype patients could also be of great influence to changes in eating behavior. Concerning the rationale for this, we hypothesized that disgust-sensitivity or food aversion in adolescents could be a classically conditioned behavior to negatively labeled physiological responses. Accordingly, physical stimuli that refer to saturation, for example, “a full stomach” (CS), would lead to a UCS/UCR representation: “I cannot stand a full stomach”, which is based on the patient’s personal learning history: “Eating in the absence of hunger leads to nausea and, when persisting, to vomiting and embarrassment.” The conditioned response (CR) to this could be panic. So, the patient will try to avoid these responses by only eating small amounts. Violating the expectancies of the UCS/UCR representation, by letting him/her gradually eat larger amounts and exposing him/her to the physical responses while the expected outcome does not occur and panic decreases, could lead to acceptance of larger amounts, and eventually to weight gain (see Figure 1).

We developed an innovative exposure based CBT treatment, by integrating the inhibitory learning principles, for adolescents with ARFID. The purpose of the current proof of concept study was to test this new treatment program. We hypothesized that ARFID symptoms would decrease in all ARFID presentations. That is, food acceptance would increase (restriction and selectivity would decrease), and associated ARFID symptoms (underweight, nutritional deficits—including dependence of supplements, and interference with daily life) would decrease, likewise. Furthermore, we expected a significant reduction in the believability of distorted cognitions about food intake, the anxiety of food and eating, and food neophobia.
2.1 Ethical approval

The present study was approved by the Ethical Research Committee of the Faculty of Psychology and Neurosciences (ERCPN) of Maastricht University, number ERCPN-164_16_03_2016.

2.2 Participants

Patients were 11 adolescents (4 females), referred by somatic specialists in child health care—who carried out the first ARFID screening—to SeysCentra, a specialized treatment facility for children with feeding disorders in the Netherlands. Their mean age was 13.9 years (SD = 1.95; range 10–17). Potential comorbid mental health conditions, such as a disorder in the autism spectrum or anxiety disorders, were not exclusion criteria. ARFID diagnosis was based on a semi-structured interview conducted at intake, including DSM-5 criteria (APA, 2013).

2.3 Procedure

All 10- to 18-year-old children on the waiting-list for SeysCentra (N = 12), were invited to participate. In order of registration, participants were divided into three treatment groups of four patients. First, adolescents and their parents were informed about the research and treatment program. If they agreed to participate in the study, they received detailed information including an informed consent. Participants could decide within 2 weeks, and finally 11 out of 12 participants signed the consent form and started treatment. One potential participant did not start treatment, eventually, resulting in group two consisting of only 3 participants.

2.4 Design

The study had a nonconcurrent multiple baseline design across subjects (Watson & Workman, 1981), constituting of an AB design (A = baseline and B = intervention). Per treatment group, each individual had a different baseline length (range: 6–9 mealtimes, over 2–3 days). Participants were randomly assigned to one of four baseline durations. After baseline, a 4-week CBT day treatment followed, continued by 4 weeks low intensity outpatient treatment. Measurements of DSM-5 ARFID diagnosis, food neophobia and related measures such as body weight and length, were taken before baseline (t1), at the end of the 4-week intensive day treatment (t2) and 3 months after the end of the 4-weeks outpatient treatment (follow-up, t3). A food selectivity test and a 1-week food diary were filled out at t1 and t3. Furthermore, continuous measurements of beliefs, anxiety, and food acceptance were taken throughout the 4-weeks intensive day treatment. Patients received 75 euros to participate in the follow-up measurement. Figure 2 displays the study design.

2.5 Treatment

2.5.1 Pre-clinical treatment phase

Before the baseline, patients engaged in two 1.5-hour “pre-clinical” sessions. In the first session, the treatment rationale was explained to both patients and parents. Furthermore, they received a food diary to track their exact food intake for seven consecutive days. Next, a food selectivity test and a 1-week food diary were filled out at t1 and t3. A food selectivity test and a 1-week food diary were filled out at t1 and t3. Furthermore, continuous measurements of beliefs, anxiety, and food acceptance were taken throughout the 4-weeks intensive day treatment. Patients received 75 euros to participate in the follow-up measurement. Figure 2 displays the study design.

2.5.2 Baseline

Eating test

A behavioral test (eating test to establish patients’ acceptance of certain foods) was carried out during baseline sessions and repeated once at follow-up. During each baseline session, at least three different food items were offered, implying a baseline varying from 18 to 27 offered food items. Patients were asked to eat these foods for breakfast, lunch, or dinner. Before presentation, the food was

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**FIGURE 1** Display of the treatment rationale regarding expectancy violation in disgust sensitivity and food aversion. Note: Violating the original CS–US expectancies occurs by gradually eating larger amounts and being exposed to the accompanying physical symptoms while the expected outcome does not occur and panic decreases; this could lead, over time, to acceptance of larger amounts, and eventually to weight gain (CS = conditioned stimulus; CR = conditioned response; US = unconditioned stimulus).
weighed by two therapists, independently. After finishing the food item (or not) the therapist and second observer independently administered the weight of the remaining food. Afterwards, the amount of consumed food was calculated as a percentage of the total, offered food. Patients were not forced or encouraged to eat the foods in the behavioral tests; the foods were just offered and patients were asked (up to three times) in a neutral manner whether they would eat these foods. At follow-up, this procedure was repeated over three sessions with three food items each (nine measures total).

### 2.5.3 | Intervention

A fundamental change to existing treatment procedures for ARFID is the application of exposure and cognitive elements. Table 1 displays an overview of treatment elements and primary interventions.

In the present study, the duration of exposure and the choice of exposed foods do not depend on decreases in anxiety or a hierarchy, respectively, but on cognitive techniques and expectancy violation. Three daily individual sessions consisted of exposure to avoided foods, behavioral experiments, cognitive restructuring, relaxation techniques, and relapse prevention. During the first stage of treatment, products were offered individually and repeatedly on a daily frequency, for a minimum of five times. Later, these were offered in combination with other (difficult) products. This also pertained to volumes, starting with a little spoonful, extending to age-adequate amounts later. Half of the foods offered during baseline measurement were practiced during treatment. Furthermore, a number of novel food items were used during treatment. At follow-up measurement, a selection of the baseline foods was offered again. About 50% of these foods were trained in the treatment and 50% were not, making it possible to investigate potential generalization effects of treatment. Besides individual sessions, patients attended one daily group session. Finally, parents were involved through weekly child–parent sessions, and by daily communicating achievements and homework.

Altogether, five CBT therapists, who received training in the protocol first, and supervision by DK and EdH, carried out all treatment sessions. Two therapists were always present, in shifts. All sessions were video recorded and randomly selected to assess treatment integrity.

The intensive 4-week day treatment was followed by a 4-week low intensity outpatient period, where patients had once or twice weekly telephone and/or live video contact. In these sessions, homework was evaluated and the individual relapse prevention program was monitored with the adolescent and his/her parent(s).

### 2.6 | Measurements

#### 2.6.1 | Food selectivity test

This test, developed by SeysCentra, determines food preferences through four colored cards stating (1) “I like that food” (green); (2) “I do not like that food” (red); (3) “I have never eaten that food” (yellow); or (4) “I would only eat that food, if...” (orange). Each patient sorted
| Session type (duration) and frequency | Pre-clinical phase | Day treatment week 1 | Day treatment week 2 | Day treatment week 3 | Day treatment week 4 | 4 week low intensity outpatient treatment |
|--------------------------------------|-------------------|----------------------|----------------------|----------------------|----------------------|-----------------------------------------|
| Individual:                          |                   |                      |                      |                      |                      |                                         |
| Investigating resistance and dysfunction | 1 Ind. session (90 min) | 14 Ind. sessions (30 min) | 14 Ind. sessions (30 min) | 14 Ind. sessions (30 min) | 14 Ind. sessions (30 min) | 4-8 Ind. sessions (30 min) |
| Determine food-item acceptance hierarchy by food selectivity test | 1 C-P session (90 min) | 5 GR. sessions (60 min) | 5 GR. sessions (60 min) | 5 GR. sessions (60 min) | 5 GR. sessions (60 min) | 2-4 C-P sessions (30 min) |
| Determine CCM’s |                    |                      |                      |                      |                      |                                         |
| Determine personal final objective |                    |                      |                      |                      |                      |                                         |
| Treatment agreement                  |                    |                      |                      |                      |                      |                                         |
| Treatment goals and elements         |                   |                      |                      |                      |                      |                                         |
| Individual:                          |                   |                      |                      |                      |                      |                                         |
| Cognitive restructuring starts       |                   |                      |                      |                      |                      |                                         |
| Seeing connections between thoughts, feelings, and behavior |                   |                      |                      |                      |                      |                                         |
| Examples of eating by using the cognitive model |                   |                      |                      |                      |                      |                                         |
| Gaining insight into cognitions and resistance |                   |                      |                      |                      |                      |                                         |
| Expanding volume and/or food items  |                   |                      |                      |                      |                      |                                         |
| Group:                               |                   |                      |                      |                      |                      |                                         |
| Sharing feeding experiences and pursuing consolidation of change |                   |                      |                      |                      |                      |                                         |
| Child–parent:                        |                   |                      |                      |                      |                      |                                         |
| Explanation of rationale and treatment procedure |                   |                      |                      |                      |                      |                                         |
| Parent and Self-monitoring of mealtimes |                   |                      |                      |                      |                      |                                         |
| |                   |                      |                      |                      |                      |                      |                                         |
| Individual:                          |                   |                      |                      |                      |                      |                                         |
| Experiencing expectancy violation after exposures and behavioral experiments |                   |                      |                      |                      |                      |                                         |
| Exercises to replace dysfunctional thoughts with functional thoughts ("helping thoughts") |                   |                      |                      |                      |                      |                                         |
| Expanding volume and/or food items  |                   |                      |                      |                      |                      |                                         |
| Group:                               |                   |                      |                      |                      |                      |                                         |
| Weekend evaluation                   |                   |                      |                      |                      |                      |                                         |
| Gaining insight in healthy nutrients |                   |                      |                      |                      |                      |                                         |
| Learning from each other’s helping thoughts |                   |                      |                      |                      |                      |                                         |
| Child–parent:                        |                   |                      |                      |                      |                      |                                         |
| Risk inventory of avoidance          |                   |                      |                      |                      |                      |                                         |
| Targets for acceptance and generalization |                   |                      |                      |                      |                      |                                         |
| Child–parent:                        |                   |                      |                      |                      |                      |                                         |
| Consolidation of changed eating behavior at home |                   |                      |                      |                      |                      |                                         |
| Risk inventory                      |                   |                      |                      |                      |                      |                                         |
| Group:                               |                   |                      |                      |                      |                      |                                         |
| Weekend evaluation                   |                   |                      |                      |                      |                      |                                         |
| Teaching relaxation in group setting |                   |                      |                      |                      |                      |                                         |
| Observing change in eating behavior in group members |                   |                      |                      |                      |                      |                                         |
| Child–parent:                        |                   |                      |                      |                      |                      |                                         |
| Relapse prevention plan              |                   |                      |                      |                      |                      |                                         |
| Generalization and outpatient plan   |                   |                      |                      |                      |                      |                                         |
| Child–parent:                        |                   |                      |                      |                      |                      |                                         |
| Relapse prevention                   |                   |                      |                      |                      |                      |                                         |
| Child–parent interaction             |                   |                      |                      |                      |                      |                                         |
### TABLE 1 (Continued)

| CBT techniques, instruments, and measures | Pre-clinical phase | Day treatment week 1 | Day treatment week 2 | Day treatment week 3 | Day treatment week 4 | 4 week low intensity outpatient treatment |
|------------------------------------------|--------------------|----------------------|----------------------|----------------------|----------------------|-----------------------------------------|
| • FST                                    | Cognitive restructuring | • Cognitive restructuring | • Cognitive restructuring | • Consolidation of changed cognitions |
| • Reinforces assessment                   | (gradual) exposure   | (gradual) exposure   | Exposure              | Exposure              |
| • FNS                                    | Behavioral experiments | Behavioral experiments | Behavioral experiments | Behavioral experiments |
| • Anthropology: (body weight/length)      | Psycho-education    | Psycho-education    | Helping thoughts      | Helping thoughts      |
| • Food diary                             | Reinforcement       | Helpine thoughts    | Relaxation            | Relaxation            |
| • Letter of intent                       | Homework exercises (self-monitoring and generalization of techniques) | Homework exercises (self-monitoring and generalization of techniques) | Homework exercises (self-monitoring and generalization of techniques) | Homework exercises (self-monitoring and generalization of techniques) |
|                                         | Self-evaluation CCM's | Self-evaluation CCM's | Self-evaluation CCM's | Self-evaluation CCM's |
|                                         | Anthropology        | Anthropology        | Anthropology          | Anthropology          |

#### 2.6.3 Food neophobia scale

The Food neophobia scale (FNS; Pliner & Hobden, 1992) is a 10-item questionnaire scored on a 7-point Likert scale, investigating the presence and severity of food neophobia—defined as the fear and avoidance of certain foods. Each item (e.g., "I do not believe this at all") can be categorized as "true" or "false" in the Food Selectivity Test. Patients scored the believability of these so-called CCMs (e.g., "I do not believe this at all") before and at the end of each exposure session on a visual analogue scale (VAS), where 0 indicates "I do not believe this at all" and 100 indicates "I believe this at all." Expectancy violation was calculated by subtracting post scores from pre-scores. Each item type was scored on a "0" (true) to "7" (false) scale, and the final composite score was obtained by adding the scores of all items. A higher score indicated a higher degree of food neophobia.

#### 2.6.4 Expectancy violation

To investigate the existence and violation of if CS, then US expectations, a continuous measurement was applied during all exposures. In the preclinical phase, therapists formulated one to three CCMs that were qualified as "true." During each exposure, the therapists asked the child to restate these expectations while the therapists reformed the CCMs as "false." This process continued until the child agreed with the therapists' formulation. The therapists verified that the child believed the CCMs were false. The therapists then asked the child to restate the CCMs as "true" and to describe the consequences of the false beliefs when eating nonpreferred food items. These measures were taken weekly. From the food diary and anthropological data, the percentage of food intake was calculated. The patients were asked to qualify their suffering in terms of "no," "sometimes," and "a lot," and to rate their suffering on a scale from 0 to 70, with scores above 35 suggesting high scores.

#### Note.

C-P = child—parent; GR. = group; Ind. = individual; CCM = causal catastrophic misinterpretation; FST = food selectivity test; FNS = food neophobia scale; VAS = visual analogue scale.

Each patient and his/her parent were interviewed at intake on the basis of a prescribed format considering the presence of ARFID criteria. It was also established whether food refusal was a consequence of lack of interest (LOI), and/or of sensory processing problems (SP), and/or of anxiety about aversive consequences (AC). The patient was asked to qualify the suffering in terms of "no," "sometimes," and "a lot," and to rate their suffering on a scale from 0 to 70, with scores above 35 suggesting high scores.

#### 2.6.2 DSM-5 ARFID Index

Each patient and his/her parent were interviewed at intake on the basis of the DSM-5 ARFID criteria. It was also established whether food refusal was a consequence of lack of interest (LOI), and/or of sensory processing problems (SP), and/or of anxiety about aversive consequences (AC). The DSM-5 ARFID index has four categories: (1) Food refusal score (FRS); (2) Food selectivity score (FSS); (3) Food avoidance score (FAS); and (4) Food Health score (FHS). The DSM-5 ARFID index is a composite score from the number of items in the "red," "orange," "yellow," and the "green" category. The DSM-5 ARFID index is a composite score from the number of items in the "red," "orange," "yellow," and the "green" category.
2.6.5 | Anxiety reduction

In patients 5–11 (group 2 and 3), anxiety was added as a second continuous measurement during exposure sessions. Before the food exposure, patients were asked to indicate their fear of eating on a VAS (‘0’ = “no anxiety”; “100” = “very anxious”). After they decided their meal was finished, fear was scored again.

2.6.6 | Food intake

For all exposure sessions, the consumed exposure food was measured in grams. Afterwards, the amount of the consumed food was calculated as a percentage of the totally offered food.

2.6.7 | Food frequency

The food diary includes information on pre-test and follow-up amounts and diversity of nutrients, the ratio between oral food intake and tube feeding, and the amount of additional medical nutrition. Calculations were done by an independent dietician.

2.7 | Data reduction and statistical analyses

First, scores of CCMs per session were averaged over pre- and post-measures at each mealtime. The course of anxiety for all meals pre-and post-intervention was conducted likewise. Fear differences were calculated per session (average fear “after” minus average fear “before”). For patient 5–11, we also calculated the correlations between expectancy violation and anxiety difference, and between both expectancy violation and anxiety difference with consumed amount of food intake. Next, we compared means over phases per patient, by calculating effect sizes. Effect sizes were computed by nonoverlap of all pairs (NAP; Parker & Vannest, 2009). Measures were computed with the Shiny app for Single-Case Data Analysis (Shiny SCDA) developed by De Kumar, Michiels, Vlaeyen, and Onghena (2018). Tables 3–5 include means over phases, and comparisons (differences scores and NAP) between baseline and treatment, treatment and follow-up, and baseline and follow-up for all patients for believability, anxiety, and food intake, respectively.

3 | RESULTS

3.1 | Patient characteristics

Table 2 shows patient characteristics and outcome measures. Follow-up measurements of patient 4 and 8 were unavailable due to their drop-out of the research (but not treatment).

3.2 | Food selectivity test

As can be seen in Table 2, food acceptance increased at t2. In 10 patients, which automatically indicates a decrease on the Food Selectivity Composite Score. This means that in 91% of the patients, food acceptance had increased while selectivity had decreased at follow-up (t3), relative to baseline measures (t1).

3.3 | DSM-5 ARFID index

Table 2 shows that each patient had ARFID, with at least two criteria at baseline (t1). At the end of treatment (t2), six patients (55%) did not meet any of the ARFID criteria anymore. In the other five patients (45%), at least two criteria disappeared. Follow-up (t3) measures show that 10 patients (91%) did not meet any ARFID criteria anymore. One (9%) patient was still underweight (SDS length for weight − 1.9).

3.4 | Food neophobia scale

Table 2 shows a decrease in scores between baseline (t1) and end of the intensive day treatment (t2) for all patients, except for patient 9 (91%). For all patients, baseline levels were in the high range (>35). At t2, eight patients (72%) remained above this, despite a general decrease in food neophobia scores. Although scores decreased further in 10 out of 11 patients, follow-up measures still showed high scores in 5 patients (45%), ranging from 37 to 47.

3.5 | Expectancy violation

Mean believability levels before and after meals in the different phases, difference of phase means for pre- and postscores and effect sizes (NAP) are outlined in Table 3. As can be seen, over all phases, expectancies were violated in all 11 patients and—for the majority—gradually declined from baseline to treatment weeks 1–4. Follow-up levels in patient 1, 7, and 11 showed a mild increase, as opposed to end of treatment levels, though expectancies were still violated compared to baseline. In the other patients, expectancies were more violated at follow-up, on average, than in the treatment weeks, except for patient 2, who already displayed low average CCMs before the start.

Regarding effect sizes, Parker, Vannest, and Davis (2011) indicate that NAP’s between 50 and 70% are questionable, between 70% and 90% effective, and above 90% highly effective. Table 3 indicates that treatment effectively decreased believability in five patients. At follow-up, this was the case for at least seven patients, of whom four had “highly effective” NAPs (two patients had no follow-up data).

3.6 | Anxiety reduction

In Table 4, the average pre- and post-intervention anxiety scores of patient 5 up to 11 are presented. Average anxiety levels decreased from baseline to intervention week 1–4, in all seven patients. In three patients, average anxiety levels substantially increased during the follow-up phase, after having decreased during treatment; particularly patients 7 and 11 increased to about halfway their baseline levels. Effect sizes indicate that treatment effectively decreased anxiety in
frequently in seven patients (in 1 "highly effectively," even). At follow-up, in at least six out of seven patients NAP corresponded with "effective treatment" (in two with "highly effective," even). One patient had no follow-up data.

3.7 | Food intake

In Table 5, the average total consumed amount of food per mealtime over baseline, day treatment, and follow-up is shown. Inspection of the means implies that the largest amounts of food were accepted during the treatment phase. In the baseline phase, accepted amounts per patient varied from 0 to 100% with a median of 0, whereas during treatment, the median per patient was 100 and a 0% score was observed only once (patient 7). In the follow-up phase, acceptance varied. The nine patients who participated in the follow-up, show an increase in the amount of accepted and eaten foods during and after intervention. Effect sizes are generally large, NAP varying from more than 90% in five patients and between 70 and 90% in the remainder 6 patients after treatment. At follow-up, five patients showed "effective" NAP scores (of whom 2 had "very effective"). Four returned to "questionable" (3 scored just below 70%), and 2 had no follow-up data.

3.8 | Food frequency

The food diary data of all 11 adolescents indicated a more varied food acceptance and an average, age-adequate nutritional intake at follow-up. Patient 2, with a restrictive eating pattern and underweight at t1 (without nutritional supplement), gained insufficient weight throughout treatment. Therefore, an additional nutritional supplement was added at t3. On the contrary, additional nutrition was completely eliminated at t3 in the six patients who were partly-dependent on tube feeding or nutritional supplements.

3.9 | Expectancy violation, anxiety reduction, and food intake correlations

The Pearson correlation between expectancy violation and anxiety reduction was \( r = 0.44 \) (p < 0.001), indicating a moderate to high association between violating expectancies and anxiety reduction. Thus, less believability of dysfunctional cognitions was associated with reduced anxiety.

The Pearson correlation between expectancy violation and food intake was \( r = 0.25 \) (p < 0.03), indicating a weak association between violating expectancies and food intake. The correlation between less anxiety and food intake was \( r = 0.16 \) (p < 0.01), indicating that less anxiety was associated with more food intake.

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DISCUSSION

Little is known about effective treatment of ARFID, especially in adolescents and adults. In this case series, we investigated whether a new exposure-based CBT led to a reduction in restrictive and/or selective eating, and to a decrease in associated symptoms such as underweight, nutritional deficits, and interference with daily life. Furthermore, we investigated the believability of dysfunctional cognitions about (the consequences of) food (intake), the anxiety related to food and eating, and extent of food neophobia. It was found that restrictive and selective eating patterns and associated ARFID symptoms decreased in all (11) patients after CBT. In 10 patients, the ARFID diagnosis was even "in remission" at follow-up. All 11 patients showed a more varied food acceptance at follow-up, which was also confirmed by the food diary data. In six patients, additional tube feeding was eliminated. One patient, still underweight at follow-up despite weight

### TABLE 3 Phase means, difference of phase means (OTS) between baseline, treatment and follow-up, and effect sizes (NAP) for "believability" before and after exposure sessions

|                 | Baseline-treatment | Treatment-follow-up | Baseline-follow-up |
|-----------------|--------------------|---------------------|--------------------|
|                 | B      | T      | F-U    | OTS    | NAP    | OTS    | NAP    | OTS    | NAP    |
| Patient 1       |        |        |        |        |        |        |        |        |        |
| Pre-exposure    | 54.92  | 59.92  | 58.42  | −5.00  | 0.49   | 1.50   | 0.50   | −3.5   | 0.46   |
| Post-exposure   | 46.58  | 44.38  | 45.75  | 2.20   | 0.50   | −1.37  | 0.50   | 0.83   | 0.46   |
| Patient 2       |        |        |        |        |        |        |        |        |        |
| Pre-exposure    | 9.47   | 9.47   | 5.55   | 0.00   | 0.46   | 3.92   | 0.66   | 3.93   | 0.61   |
| Post-exposure   | 9.84   | 7.62   | 5.40   | 2.22   | 0.55   | 2.63   | 0.69   | 4.44   | 0.65   |
| Patient 3       |        |        |        |        |        |        |        |        |        |
| Pre-exposure    | 51.19  | 37.25  | 3.15   | 13.94  | 0.65   | 34.10  | 0.91   | 48.04  | 1.00   |
| Post-exposure   | 49.53  | 31.34  | 2.05   | 18.19  | 0.69   | 29.29  | 0.88   | 47.48  | 1.00   |
| Patient 4       |        |        |        |        |        |        |        |        |        |
| Pre-exposure    | 43.61  | 62.78  | −19.16 | 0.25   |         |        |        |        |        |
| Post-exposure   | 24.28  | 42.15  | −17.87 | 0.28   |         |        |        |        |        |
| Patient 5       |        |        |        |        |        |        |        |        |        |
| Pre-exposure    | 27.56  | 10.46  | 0.26   | 17.11  | 0.64   | 10.20  | 0.83   | 27.30  | 0.79   |
| Post-exposure   | 25.10  | 6.43   | 0.00   | 18.67  | 0.65   | 6.43   | 0.84   | 25.10  | 0.84   |
| Patient 6       |        |        |        |        |        |        |        |        |        |
| Pre-exposure    | 54.15  | 44.39  | 0.11   | 9.76   | 0.70   | 44.28  | 1.00   | 54.04  | 0.99   |
| Post-exposure   | 48.15  | 35.86  | 0.33   | 12.29  | 0.66   | 35.53  | 1.00   | 47.82  | 0.96   |
| Patient 7       |        |        |        |        |        |        |        |        |        |
| Pre-exposure    | 65.67  | 13.20  | 16.60  | 52.46  | 0.85   | −3.40  | 0.42   | 49.07  | 0.83   |
| Post-exposure   | 65.96  | 7.25   | 13.80  | 58.71  | 0.98   | −6.55  | 0.42   | 52.16  | 0.86   |
| Patient 8       |        |        |        |        |        |        |        |        |        |
| Pre-exposure    | 31.17  | 25.04  | 6.13   | 0.62   |         |        |        |        |        |
| Post-exposure   | 47.78  | 34.3   | 13.44  | 0.69   |         |        |        |        |        |
| Patient 9       |        |        |        |        |        |        |        |        |        |
| Pre-exposure    | 50.36  | 23.53  | 10.47  | 26.82  | 0.82   | 13.07  | 0.82   | 39.89  | 0.90   |
| Post-exposure   | 48.07  | 19.56  | 5.63   | 28.50  | 0.81   | 13.93  | 0.82   | 42.44  | 0.93   |
| Patient 10      |        |        |        |        |        |        |        |        |        |
| Pre-exposure    | 96.16  | 47.24  | 6.00   | 48.97  | 0.86   | 41.24  | 0.86   | 90.21  | 1.00   |
| Post-exposure   | 97.63  | 42.72  | 6.50   | 54.91  | 0.86   | 36.22  | 0.80   | 91.13  | 1.00   |
| Patient 11      |        |        |        |        |        |        |        |        |        |
| Pre-exposure    | 68.62  | 11.37  | 38.68  | 57.25  | 0.90   | −27.31 | 0.29   | 29.94  | 0.75   |
| Post-exposure   | 70.90  | 7.86   | 50.36  | 62.98  | 0.93   | −42.44 | 0.20   | 20.54  | 0.71   |

Note. NAP: Percentage Non-overlap of All Pairs. NAP equals the number of comparison pairs showing no overlap, divided by the total number of comparisons. We obtained NAP by using the Shiny app for Single-Case Data Analysis (Shiny SCDA), provided by KU Leuven (Kumar De, T., Michiels, B., Vlaeyen, J.W.S., & Onghena, P.).

OTS: Observed Test Statistic: mean difference between the several phases. Positive numbers indicate a decrease, negative numbers an increase (of believability). B, Baseline; T, Treatment; F-U, Follow-up.
improvement, was advised an additional medical nutritional supplement. Psychosocial interference improved, as well: all adolescents evaluated their treatment as positive and indicated “less interference with psychosocial functioning, as compared to baseline.” Food neophobia ratings for all adolescents decreased from high levels to nearby or below 35.

Next to objective ARFID symptoms, we investigated violation of negative expectations and the course of anxiety during baseline, treatment, and follow-up. In all patients, expectancies were violated and, in line with that, anxiety levels pre-post exposure sessions decreased during treatment. At 3-months follow-up, these results were retained in six patients, while believability of dysfunctional cognitions and anxiety levels increased somewhat again in three patients (although these remained below baseline levels). Thus, we found that most patients showed less anxiety and were able to adjust their dysfunctional beliefs after exposure.

Furthermore, we investigated whether expectancy violation was associated with anxiety reduction and whether either of the two was (to a greater extent) associated with food intake. The correlations indeed support relations between anxiety reduction and expectancy violation, and both were associated with food intake. Although these data do not allow any conclusions about causality, the fact that both mechanisms (expectancy violation and fear reduction/habituation) appear to be related to an improvement in ARFID symptoms, is interesting. In this respect, we could question whether we should focus the food exposure on fear reduction or on expectancy violation (or both?). Similar results were found by Schyns, Roefs, et al. (2018), who measured both craving (reduction) and expectancy violation during cue-exposure treatment for overeating. Both procedures (habituation and expectancy violation) led to less eating desires and a decreased food intake at post-test, but neither was found more effective and could be assigned as a significant mediator. It could be hypothesized that the underlying learning mechanism is individually determined; that is, for one patient habitual learning might be the better moderator whereas for another patient expectancy violation is a more powerful means. Further research should be conducted regarding potential individual differences or sensitivity towards both mechanisms. For the time being, we suggest to implement both procedures (expectancy violation and habitual learning) in the exposure treatment for ARFID, in order to generate a maximum effect.

Our sample included two patients (7 and 11) with a comorbid generalized anxiety disorder. Since we observed a substantial increase in

| TABLE 4 | Phase means, difference of phases means (OTS) between baseline, treatment and follow-up, and effect sizes (NAP) for “anxiety” before and after exposure sessions |
|---------|-------------------------------|-----------------|-----------------|-----------------|-----------------|
|         | Means                         | Baseline-treatment | Treatment-follow-up | Baseline-follow-up |
|         | B    | T    | F-U  | OTS   | NAP  | OTS   | NAP  | OTS   | NAP  |
| Patient 5 |      |      |      |       |      |       |      |       |      |
| Pre-exposure | 16.75 | 12.40 | 0.22 | 4.35  | 0.53 | 12.17 | 0.84 | 16.53 | 0.82 |
| Post-exposure | 15.75 | 6.23  | 0.00 | 9.52  | 0.61 | 6.23  | 0.80 | 15.75 | 0.81 |
| Patient 6 |      |      |      |       |      |       |      |       |      |
| Pre-exposure | 48.90 | 44.23 | 1.22 | 4.67  | 0.65 | 43.01 | 0.96 | 47.68 | 0.95 |
| Post-exposure | 41.10 | 29.02 | 0.98 | 12.08 | 0.61 | 28.13 | 0.93 | 40.21 | 0.92 |
| Patient 7 |      |      |      |       |      |       |      |       |      |
| Pre-exposure | 72.17 | 16.43 | 34.70 | 55.73 | 0.87 | −18.27 | 0.34 | 37.47 | 0.80 |
| Post-exposure | 66.54 | 9.89  | 28.80 | 56.66 | 0.86 | −18.91 | 0.41 | 37.74 | 0.75 |
| Patient 8 |      |      |      |       |      |       |      |       |      |
| Pre-exposure | 69.42 | 43.88 | 0.90 | 25.54 | 0.81 |       |      |       |      |
| Post-exposure | 51.00 | 29.06 | 0.66 |       |      |       |      |       |      |
| Patient 9 |      |      |      |       |      |       |      |       |      |
| Pre-exposure | 78.07 | 54.18 | 23.29 | 23.89 | 0.80 | 30.98 | 0.81 | 54.87 | 0.95 |
| Post-exposure | 72.60 | 36.50 | 18.10 | 36.10 | 0.86 | 18.40 | 0.72 | 54.50 | 0.90 |
| Patient 10 |      |      |      |       |      |       |      |       |      |
| Pre-exposure | 73.58 | 15.98 | 29.00 | 57.60 | 0.85 | −13.02 | 0.28 | 44.58 | 0.83 |
| Post-exposure | 83.47 | 12.10 | 34.58 | 71.37 | 0.91 | −22.48 | 0.25 | 48.89 | 0.89 |
| Patient 11 |      |      |      |       |      |       |      |       |      |
| Pre-exposure | 69.26 | 15.00 | 40.38 | 54.26 | 0.90 | −20.18 | 0.29 | 28.88 | 0.75 |
| Post-exposure | 68.62 | 7.86  | 49.91 | 60.76 | 0.93 | −42.05 | 0.14 | 18.71 | 0.67 |

NAP: Percentage Non-overlap of All Pairs. NAP equals the number of comparison pairs showing no overlap, divided by the total number of comparisons. We obtained NAP by using the Shiny app for Single-Case Data Analysis (Shiny SCDA), provided by KU Leuven (Kumar De, T., Michiels, B., Vlaeyen, J.W.S., & Onghena, P.).

OTS: Observed Test Statistic: mean difference between the several phases. Positive numbers indicate a decrease, negative numbers an increase (of anxiety). B, Baseline; T, Treatment; F-U, Follow-up.
anxiety as well as a decrease in expectancy violation at follow-up in these patients, in contrast to the other patients (without anxiety diagnoses), it could be that patients with a comorbid anxiety disorder are more at risk for relapse. Future research might include the relative effects of treatment when several types of comorbidities are present. This was also the case in the two LOI patients where, on average, lower rates were observed regarding believability as well as anxiety. It might be hypothesized that individuals with less appetite profit more from the habitual learning paradigm than from inhibitory learning. Given the small sample size, generalized conclusions are premature and future research on particular subtypes should show whether this hypothesis remains valid.

Two patients were lost to follow-up evaluations. We were, however, able to obtain questionnaire data from both, providing some information on their eating at follow-up. Furthermore, this study was a clinical case series with only 11 patients. Still, the results of this proof of concept study, whose primary aim was to detect whether a new type of CBT leads to a decrease in ARFID symptoms, are quite promising. Larger (RCT) studies including more patients with various comorbidities and different ARFID presentations are necessary, to find out more about CBT’s effectiveness and secondly, to study whether adapted (longer? more intense?) CBT should be applied in different ARFID subtypes.

We developed a short intensive day-treatment. Unfortunately, data on in- or outpatient ARFID treatment in the Netherlands are not available. In general, patients with anorexia nervosa are more often treated in a hospitalized setting or day treatment than in outpatient settings. We believe that for ARFID, more intensive (day) treatment is justified as the target problem (eating) occurs several times a day; therefore, treatment should be carried out on as many occasions as possible to acquire a relatively fast change.

In conclusion, this case series suggests that our CBT protocol was effective in reducing ARFID symptoms in 10- to 18-year-old adolescents. Further research is necessary to study what the causal mechanisms (such as expectancy violation or habituation) exactly are.

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| TABLE 5  | Phase means, difference of phases means (OTS) between baseline, treatment and follow-up, and effect sizes (NAP) for percentage of food intake during exposure sessions |
|----------|-------------------------------------------------------------------------------------------------------------------------------|
|          | Means | Baseline-treatment | Treatment-follow-up | Baseline-follow-up |
|          | B     | T     | F-U | OTS   | NAP | OTS   | NAP | OTS   | NAP |
| Patient 1|       |       |     |       |     |       |     |       |     |
| Post-exposure | 51.1 | 94.29 | 54.90 | 43.17 | 0.79 | −39.34 | 0.17 | 3.82 | 0.53 |
| Patient 2|       |       |     |       |     |       |     |       |     |
| Post-exposure | 43.00 | 84.62 | 90.00 | 41.60 | 0.81 | 5.37 | 0.46 | 46.98 | 0.85 |
| Patient 3|       |       |     |       |     |       |     |       |     |
| Post-exposure | 7.64 | 100.00 | 33.90 | 92.36 | 0.97 | −66.07 | 0.05 | 26.30 | 0.92 |
| Patient 4|       |       |     |       |     |       |     |       |     |
| Post-exposure | 17.70 | 81.68 | 64.03 | 0.92 |     |       |     |       |     |
| Patient 5|       |       |     |       |     |       |     |       |     |
| Post-exposure | 36.80 | 98.77 | 93.40 | 62.02 | 0.84 | −5.33 | 0.45 | −56.69 | 0.82 |
| Patient 6|       |       |     |       |     |       |     |       |     |
| Post-exposure | 76.50 | 99.91 | 97.00 | 23.41 | 0.69 | −2.91 | 0.45 | 20.51 | 0.95 |
| Patient 7|       |       |     |       |     |       |     |       |     |
| Post-exposure | 30.40 | 89.26 | 50.60 | 58.88 | 0.85 | −38.70 | 0.26 | 20.18 | 0.64 |
| Patient 8|       |       |     |       |     |       |     |       |     |
| Post-exposure | 33.30 | 96.39 | 63.13 | 0.86 |     |       |     |       |     |
| Patient 9|       |       |     |       |     |       |     |       |     |
| Post-exposure | 22.80 | 97.58 | 64.60 | 74.81 | 0.95 | −32.98 | 0.22 | 41.83 | 0.84 |
| Patient 10|       |       |     |       |     |       |     |       |     |
| Post-exposure | 5.72 | 9.89 | 57.00 | 87.17 | 0.97 | −35.84 | 0.20 | 51.32 | 0.95 |
| Patient 11|       |       |     |       |     |       |     |       |     |
| Post-exposure | 8.02 | 97.98 | 30.10 | 89.95 | 1.00 | −67.83 | 0.10 | 22.12 | 0.64 |

NAP: Percentage Non-overlap of All Pairs. NAP equals the number of comparison pairs showing no overlap, divided by the total number of comparisons. We obtained NAP by using the Shiny app for Single-Case Data Analysis (Shiny SCDA), provided by KU Leuven (Kumar De, T., Michiels, B., Vlaeyen, J.W.S., & Onghena, P.).

OTS: Observed Test Statistic: mean difference between the several phases. Positive numbers indicate an increase, negative numbers a decrease (of food intake). B, Baseline; T, Treatment; F-U, Follow-up.
sizes within Single Case Experimental Designs and for providing the “Shiny app for Single-Case Data Analysis (SCDA).”

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