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BRIEF REPORT

Exploring the cooccurrence of behavioural phenotypes for avoidant/restrictive food intake disorder in a partial hospitalization sample

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Abstract

Objective: Literature providing clinical characterizations of avoidant/restrictive food intake disorder (ARFID) has proposed the occurrence of three functions for food refusal: fear of negative consequences, lack of hunger, or sensory sensitivity. Recent studies have suggested that these functions may be used to subtype patients presenting with ARFID; however, other work suggests that these categories are not mutually exclusive and instead represent neurobiological dimensions that can cooccur. The current study explored the potential cooccurrence of behavioural phenotypes in patients with ARFID presenting to a partial hospitalization program.

Method: Two raters conducted a retrospective chart review of patients with ARFID presenting to treatment from June 2014 to May 2018 ($N = 59$).

Results: Regarding cooccurrence of symptoms consistent with behavioural phenotypes, raters showed excellent agreement, and over 50% of the sample endorsed symptoms consistent with more than one phenotype. The sensory sensitivity phenotype was most common in the sample and frequently cooccurred with both other phenotypes.

Discussion: Results suggest that multiple functions for food avoidance may be present within one individual. Future work should aim to further characterize individuals presenting with singular versus multiple phenotype characteristics.

KEYWORDS
appetite, avoidant/restrictive food intake disorder, eating disorder, negative affect, selective eating

1 | INTRODUCTION

Avoidant/restrictive food intake disorder (ARFID) is the newest eating disorder in the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5; American Psychiatric Association, 2013). ARFID captures a heterogeneous collection of restrictive eating behaviours that result in weight loss, poor growth, nutritional deficiency, dependence on oral or enteral supplements, or psychosocial impairment. Currently, the DSM-5 lists three characteristic functions of restrictive eating: (1) dietary restriction secondary to sensory sensitivity, (2) fear of aversive consequences while eating, and (3) lack of interest in eating secondary to poor appetite. Clinicians and researchers have further recognized the diversity of presentations among patients with ARFID, and clinical observations suggest that these three functions of food refusal may represent clinically meaningful behavioural

phenotypes (Fisher et al., 2014; Kennedy, Wick, & Keel, 2018; Norris et al., 2018; Thomas et al., 2017). Considering data suggesting that patients with ARFID may be more likely to drop out of treatment (Forman et al., 2014), exploring empirical methods for understanding symptom heterogeneity may offer one approach to more effectively tailor psychological treatments and promote better outcomes.

To date, some literature has taken the approach of using distinct functions for food avoidance as a way to classify individuals with ARFID into subtypes; in the DSM-5, diagnostic subtypes are more generally defined as "mutually exclusive and jointly exhaustive phenomenological subgroupings within a diagnosis" (American Psychiatric Association, 2013). Although referred to by different names by different groups, the proposed groups were initially characterized by Bryant-Waugh, Markham, Kreipe, and Walsh (2010) and reflect the three main restrictive eating functions characterized in the DSM-5's description of Criterion A for ARFID. Although empirical data exploring distinct subtypes of ARFID remains limited, one recent study indicated that the proposed subtypes could be identified with moderate inter-rater reliability and demonstrated differences with regard to length of illness and hospitalization history (Norris et al., 2018). Subtype classifications from this investigation suggested that the fear of aversive consequences and poor appetite subtypes were most common in a treatment-seeking sample. Moreover, a recent investigation of children presenting to a paediatric gastroenterology health care network indicated that of the identified ARFID cases in the sample, the majority fit the low appetite subtype (~58%), with fewer endorsing symptoms consistent with the sensory sensitivity (~21%) and fear of aversive consequences (~9%) subtypes (Eddy et al., 2015).

Although some researchers have explored the existence of distinct subtypes of ARFID, another approach considers differential food functions as behavioural phenotypes that have distinct neurobiological aetiologies and can coexist within an individual (Thomas et al., 2017). Clinicians and researchers that endorse this approach argue that patients with ARFID often present for treatment with restrictive eating behaviours that fall into multiple functional domains. The proposed domains reflect impairment in sensory perception (i.e., sensory sensitivity), negative valence systems (i.e., fear of aversive consequences), and homeostatic appetite regulation (i.e., low appetite; Thomas et al., 2017). Several recent studies in large community samples of adult and youth picky eaters (Kurz, van Dyck, Dremmel, Munsch, & Hilbert, 2016; Wildes, Zucker, & Marcus, 2012) have suggested that differing food avoidance functions can cooccur, although the criteria for assessing different picky eating characteristics in these studies varied and did not include explicit assessment of the poor appetite symptoms. To date, no studies have explored the frequency with which characteristics of proposed behavioural phenotypes overlap, particularly in a treatment-seeking sample. Accordingly, the current study aims to provide a preliminary investigation of the frequency of various behavioural phenotypes in a sample of patients presenting for treatment at a day treatment program for paediatric eating disorders.

2 | METHODS

2.1 | Patient population

Data were collected as part of a retrospective chart review of patients enrolled in a paediatric program of an eating disorders treatment and research facility between the commencement of the program in June 2014 and May 2018. Inclusion criteria for the study were a primary diagnosis of ARFID. Diagnoses were first assigned by a masters-level clinician at an intake interview, which were then confirmed by an unstructured clinical interview completed by psychiatrists during the patient's first week at the clinic. In the event that there were discrepancies between the intake and psychiatrist diagnosis, these were resolved by a consensus meeting held by the patient's treatment team (psychiatrist, family therapist, and individual therapist). There were no exclusion criteria for the study. If patients admitted to the clinic more than one time ($n = 4$), data from their first enrolment were coded for inclusion in the study. The following information was gathered from each chart: gender, age at intake, age of onset for feeding difficulties/duration of illness, racial/ethnic background, body weight and height at intake to treatment, percent of ideal body weight (based on individualized growth curves), and medications at intake. We also gathered information on medical and psychiatric comorbidities as assessed and diagnosed by program psychiatrists.

2.2 | Coding of clinical characteristics

Author J. E. M. developed a hierarchical coding system based on prior literature describing features of proposed symptom presentation (Bryant-Waugh et al., 2010; Cooney, Lieberman, Guimond, & Katzman, 2018; Norris et al., 2018; Thomas et al., 2017), as well as clinical expertise; the coding sheet is available in Table S1 in the Supporting Information. Clinical characteristics were then coded by two doctoral-level individual raters (E. E. R. and T. A. B.) that had no prior familiarity with the patients in the paediatric program. Using information in the clinical charts, including intake and discharge

summaries, clinical notes from psychiatrists, individual and family therapists, and nutritionists, and collateral information, raters coded each group of characteristics as *present* or *absent*. If symptoms were ambiguous and/or listed only one time in the chart (e.g., mention of low hunger cues, but only in one report), they were coded as absent. Of note, the proposed behavioural phenotypes are not used currently as a diagnostic tool in the clinic. Therefore, explicit mention of phenotype characteristics was not present in any charts, and raters instead used descriptive report-based information regarding the nature of food avoidance in coding. Inter-rater reliability for phenotype calculations was calculated using Cohen's κ statistic. The protocol received approval from the institution's Institutional Review Board.

2.3 | Description of program

The program in question contains both a 5-day, 6-hr partial hospitalization program and a 3- to 5-day, 3-hr intensive outpatient program and will be jointly referred to as the day treatment program throughout the remainder of this paper. Enrolment criteria for the day treatment program include being a child with a primary diagnosis of an eating disorder who is also medically stable and endorses a level of symptom severity that is not appropriate for outpatient care and warrants additional supervision (e.g., significantly low body weight and previous failure of outpatient care). Patients are excluded from participation in the program if they endorse a significant developmental disability that would prevent them from benefitting from and/or participating in a group-based therapeutic environment. Generally, the day treatment program treats children aged 6–12. Young adolescents (e.g., ages 13 or 14) are considered for enrolment in the paediatric program on a case-by-case basis that includes consideration of developmental level and diagnosis. All patients first enter the partial hospitalization program and are stepped down to intensive outpatient program care based on progress toward treatment goals. The program provides group-based, individual, and family therapy for all patients in the program, alongside dietary therapy and psychiatric medical management. The primary interventions utilized by the day treatment program include family-based treatment and cognitive behavioural therapy.

3 | RESULTS

3.1 | Demographic statistics of the sample

Overall, 133 participants admitted to the day treatment program in the specified time frame. Around 44% of the total enrolled sample was assigned a primary diagnosis of ARFID ($N = 59$) and comprised the current sample. The remainder of the patients admitted to the day treatment program were assigned other eating disorder diagnoses; the most common other diagnosis was Anorexia Nervosa, followed by Other Specified Feeding and Eating Disorder, and Rumination Disorder. No other diagnoses were represented. The sample was equally split across gender groups, predominantly white, and around 10 years old at the time of enrolment, with an average duration of illness around 34 months. However, there was significant variability in the age of onset across the sample, with 5 (8.9%) reporting feeding difficulties beginning in infancy (<12 months old), 7 (12.5%) in toddlerhood (12–36 months), and 44 (78.6%) in childhood (36 months+), with age of onset not available for 3 patients (5.1%). Around 49% of the sample was classified as underweight, as defined by a weight less than 85% of expected body weight. A variety of psychiatric and medical comorbidities were represented. As noted in Table 1, the most common co-occurring diagnoses included ADHD, GAD, Other Specified Anxiety Disorder, OCD, and Social Phobia. In terms of medical comorbidities, Crohn's Disease, Failure to Thrive, and Functional Gastrointestinal Disorders were most common.

3.2 | Frequency of ARFID behavioural phenotypes across raters

Table 1 presents descriptive statistics across individuals demonstrating the three behavioural phenotypes. The average frequency of ARFID behavioural phenotypes by gender across raters is available in Figure 1, and in Table S2, we offer a table providing the frequencies of proposed behavioural phenotypes for both raters. Finally, we also present demographic information for individuals coded as demonstrating one, two, or three phenotypes in Table S3. Cohen's κ statistics for the lack of appetite, fear of aversive consequences, and sensory sensitivity phenotypes were 0.96, 0.93, and 0.85, respectively. Of note, κ values can range from −1 to 1, and values above 0.60 indicating moderate agreement, and values over 0.80 suggesting substantial agreement (Shrout, 1998). Agreement in the rated cooccurrence across phenotypes was also excellent, Cohen's $\kappa = 0.87$. Characteristics consistent with the sensory sensitivity behavioural phenotype were most commonly observed in the sample, with between 62% and 80% of individuals reporting at least one characteristic of this phenotype. In individuals that only endorsed symptoms consistent with one behavioural phenotype, symptoms consistent with the fear of aversive consequences

TABLE 1 Demographic information by phenotype, across raters

	Selective eating *M* (*SD*)/*n* (%)		Fear of aversive consequences *M* (*SD*)/n (%)		Low appetite *M* (*SD*)/*n* (%)	
Variable	**Rater 1**	**Rater 2**	**Rater 1**	**Rater 2**	**Rater 1**	**Rater 2**
Age	10.23 (2.28)	10.31 (2.34)	10.33 (2.18)	10.26 (2.15)	10.68 (2.44)	11.05 (2.44)
%IBW at intake	85.40 (7.04)	86.34 (8.42)	86.8% (8.45)	87.1 (8.55)	82.83% (5.20)	82.40% (5.23)
Height (inches)	54.41 (4.82)	54.52 (4.96)	54.60 (4.50)	54.72 (4.61)	54.28 (6.18)	54.93 (6.05)
Weight (pounds)	65.05 (15.71)	65.95 (17.02)	66.31 (16.54)	66.52 (16.44)	62.59 (15.12)	64.25 (14.90)
Length of illness (months)	37.14 (37.42)	38.78 (37.69)	27.83 (34.92)	25.83 (30.85)	49.20 (40.90)	54.26 (44.63)
Gender (Male)	24 (54.5%)	24 (57.1%)	16 (44.4%)	16 (42.1%)	12 (54.5%)	13 (65.0%)
Racial/ethnic background						
White	34 (77.3%)	33 (78.6%)	26 (72.2%)	28 (73.7%)	19 (86.4%)	17 (85.0%)
Latinx/Hispanic	8 (18.2%)	6 (14.3%)	8 (22.2%)	8 (21.1%)	2 (9.1%)	2 (10.0%)
Asian	0 (0.0%)	1 (14.3%)	1 (2.8%)	1 (2.6%)	0 (0.0%)	0 (0.0%)
Other	2 (4.5%)	2 (4.8%)	1 (2.8%)	1 (2.6%)	1 (4.5%)	1 (5.0%)
Medical comorbidities						
Failure to thrive	6 (13.6%)	6 (14.3%)	4 (11.1%)	3 (7.9%)	6 (27.3%)	6 (30.0%)
Other medical comorbidity	5 (11.4%)	4 (9.5%)	5 (13.9%)	6 (15.8%)	2 (9.1%)	2 (10.0%)
Functional gastrointestinal disorder	1 (2.3%)	1 (2.4%)	2 (5.6%)	2 (5.3%)	0 (0.0%)	0 (0.0%)
Crohn's disease	2 (4.5%)	2 (4.8%)	0 (0.0%)	1 (2.6%)	2 (9.1%)	1 (5.0%)
Comorbid psychiatric disorders						
Attention-deficit/hyperactivity disorder	11 (25.0%)	10 (23.8%)	5 (13.9%)	4 (10.5%)	8 (36.4%)	8 (40.0%)
Generalized anxiety disorder	7 (15.9%)	8 (19.0%)	11 (30.6%)	11 (28.9%)	1 (4.5%)	1 (5.0%)
Other specified anxiety disorder	11 (25.0%)	8 (19.0%)	6 (16.7%)	7 (18.4%)	5 (22.7%)	3 (15.0%)
Obsessive compulsive disorder	3 (6.8%)	2 (4.8%)	4 (11.1%)	4 (10.5%)	0 (0.0%)	1 (5.0%)
Autism spectrum disorder	2 (4.5%)	2 (4.8%)	3 (8.3%)	2 (5.3%)	1 (4.5%)	2 (10.0%)
Social anxiety disorder	2 (4.5%)	2 (4.8%)	2 (5.6%)	2 (5.3%)	1 (4.5%)	1 (5.0%)
Separation anxiety disorder	1 (2.3%)	1 (2.4%)	1 (2.8%)	1 (2.6%)	0 (0.0%)	0 (0.0%)
Other specified depressive disorder	2 (4.5%)	2 (4.8%)	1 (2.8%)	1 (2.6%)	0 (0.0%)	0 (0.0%)
Medications at intake						
Antidepressant	7 (15.9%)	8 (19.0%)	7 (19.4%)	8 (21.1%)	4 (18.2%)	3 (15.0%)
Atypical antipsychotic	3 (6.8%)	2 (4.8%)	4 (11.1%)	3 (7.9%)	2 (9.1%)	2 (10.0%)
Mood stabilizer	1 (2.3%)	1 (2.4%)	1 (2.8%)	1 (2.6%)	1 (4.5%)	1 (5.0%)
Anxiolytic	1 (2.3%)	1 (2.4%)	2 (5.6%)	2 (5.3%)	0 (0.0%)	0 (0.0%)
ADHD medication	7 (15.9%)	7 (16.7%)	2 (5.6%)	1 (2.6%)	7 (31.8%)	8 (40.0%)

Note. %IBW: percent of ideal body weight; ADHD: attention-deficit/hyperactivity disorder.

phenotype were most commonly observed. Of note, the majority of the sample (54.2% across both raters) endorsed symptoms characteristics of multiple ARFID behavioural phenotypes. The most common cooccurrences of behavioural phenotypes were (1) fear of aversive consequences-sensory sensitivity and (2) low appetite-sensory sensitivity. Up to 10% of patients endorsed clinical symptoms consistent with all three proposed phenotypes.

4 | DISCUSSION

The present investigation represents the first empirical attempt to explore the general cooccurrence of behavioural phenotypes commonly observed in individuals with ARFID. Results from the current study indicate that over half of patients diagnosed with ARFID endorse symptoms characteristic of more than one proposed behavioural phenotype. These data suggest that a categorical classification

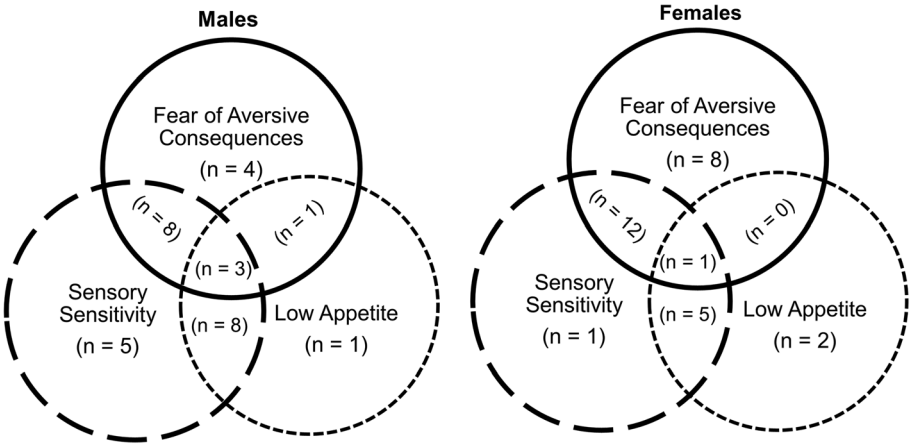

FIGURE 1 Overlap between ARFID subtypes, averaged across raters

system may not sufficiently capture symptom heterogeneity observed within a treatment-seeking paediatric sample and suggests that future research exploring different methods for classifying and assessing differential behavioural phenotypes is warranted.

Chart ratings completed by two raters demonstrated consistently high reliability; in our sample, behavioural characteristics associated with alterations in sensory sensitivity were most common in the sample, followed by fear of aversive consequences. Consistent with these findings, the cooccurrence of symptoms associated with these phenotypes was most common. The high prevalence of self-reported alterations in sensory sensitivity difficulties is consistent with past investigations of community-based samples, both of which reported food avoidance linked to sensory-based characteristics of food as present in over half of picky eaters (Wildes et al., 2012). In addition, although we did not evaluate group differences statistically due to lack of power, stratifying ratings by gender suggested that the characteristics of the low appetite phenotype were more often observed in males, whereas fear-driven avoidance of food was potentially more common in females. Higher occurrence of alterations in low appetite phenotype among male youth with ARFID is consistent with one prior study, which indicated that in a predominantly male sample of ARFID patients presenting to a gastroenterology health network, the low appetite phenotype was most common (Eddy et al., 2015). Moreover, higher frequency of food avoidance linked to fear of aversive consequences is consistent with work documenting more frequent anxiety-based psychopathology in females (Simonoff et al., 1997). Overall, our findings are supportive of past suggestions that multiple phenotypes can cooccur within an individual. However, it is important to highlight that our findings may have been secondary to the classification scheme and threshold that we set for the behavioural phenotype assessment; because there are no measurements or comprehensive definitions for ARFID behavioural phenotypes currently published in the literature, we developed an exploratory assessment scheme and a minimally restrictive threshold as a first step in answering questions related to phenotype cooccurrence. It will be critical for future work to focus on developing reliable, valid assessments of these phenotypes and more explicitly exploring thresholds for determining the presence or absence of a given phenotype. The forthcoming Pica, ARFID, and Rumination Disorder Interview (Bryant-Waugh et al., 2016) represents a promising new tool for critical work, as the assessment allows the user to generate symptom composites related to each behavioural phenotype of ARFID.

In addition to providing initial support for the cooccurrence behavioural phenotypes, data from our sample differed from previous demographic investigations of ARFID in several ways. First, this study reflects a more recent cohort of patients who presented for treatment after the introduction of ARFID into the DSM-5 nomenclature. Second, this study included a significantly higher percentage of males in a treatment-seeking sample compared with previously published samples, although one previous study conducted in a paediatric gastroenterology network also found a preponderance of males (Eddy et al., 2015). Previous studies have also found a higher proportion of males diagnosed with ARFID compared with males diagnosed with anorexia (e.g., Fisher et al., 2014; Ornstein, Essayli, Nicely, Masciulli, & Lane-Loney, 2017). However, our sample suggests that the proportion of males with ARFID may be even higher than previously speculated. Finally, this study adds to the existing literature on psychiatric comorbidities, confirming high rates of comorbid anxiety disorders, but also finding a higher than previously-reported comorbidity with ADHD. Given that prescribed psychotropic medication for ADHD has documented effects on appetite and some and initial case

reports have proposed a connection between stimulant use and ARFID symptoms (Pennell, Couturier, Grant, & Johnson, 2016), it will be important for future research to longitudinally explore the role of ADHD symptoms and prescribed medication in the onset or exacerbation of ARFID symptoms. In our sample, individuals taking stimulant mediation at intake were more likely to endorse characteristics of the low appetite phenotype, but there was only participant (12%) that *only* demonstrated low appetite-related symptoms; the rest of the individuals prescribed stimulants endorsed symptoms characteristics of other phenotypes. Overall, it is likely that the links between ADHD, stimulant use, and appetite are complex and should be a critical area for future investigation.

Although the current investigation represents an initial examination of proposed behavioural phenotype overlap, future work in this domain will have significant implications for the understanding and treatment of ARFID. Past theoretical work has suggested that treatment approaches for ARFID may be most effectively tailored to differing symptom presentations, such that specific, distinct therapeutic techniques may be warranted depending on the function of food avoidance (Bryant-Waugh et al., 2010; Thomas et al., 2017). As noted above, given our initial evidence suggesting that multiple functions for food avoidance may co-exist in an individual, it will be critical to develop streamlined assessments that can provide reliable and valid estimates of commonly-observed ARFID characteristics, as well as comprehensive treatments that can be tailored to the specific symptom profile of an individual.

4.1 | Strengths and limitations

Strengths of the current study include the use of two independent raters and inter-rater reliability statistics for the chart review protocol, as well as the consideration of a range of clinical characteristics, rather than dichotomous coding of functions for food avoidance. Limitations include the retrospective nature of data collection and a small sample, which limits generalizability of the findings and provides restricted power to evaluate more specific group differences among individuals endorsing differing co-occurring behavioural phenotypes. Finally, due to the lack of available assessment of differing ARFID phenotypes, we developed our own assessment for the purposes of chart review, and, given the exploratory nature of the study, chose to enact a low-threshold for marking a given behavioural phenotype as present. It will be critical for future work to explicitly define and develop well-validated assessments probing differing clinical features of proposed food avoidance functions.

5 | CONCLUSIONS

Despite an increase in interest regarding the clinical characteristics of ARFID, data-driven investigation of proposed behavioural phenotypes of the disorder remain limited. Results from a retrospective chart review suggest that the majority of individuals presenting to a specialized eating disorder program meet criteria for multiple proposed behavioural phenotypes of ARFID previously proposed in the literature, and demographics for the present sample differed from prior samples described in the literature along several dimensions, including gender and psychiatric comorbidities. Overall, findings support recent proposals that behavioural phenotypes in ARFID cooccur and add to the burgeoning literature seeking to better describe the clinical characteristics of the disorder.

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SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section at the end of the article.

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