OPEN

Toward Better Understanding of Pediatric Feeding Disorder: A Proposed Framework for Patient Characterization

*†William G. Sharp, PhD, ‡Alan Silverman, PhD, ‡Joan C. Arvedson, PhD,
§‖Nancy F. Bandstra, PhD, ¶Elizabeth Clawson, PhD, †Rashelle C. Berry, MPH, MS, RD,
*†Barbara O. McElhanon, MD, #**Alison M. Kozlowski, PhD, BCBA-D, ††Mitchell Katz, MD,
*†Valerie M. Volkert, PhD, BCBA-D, ‡Praveen S. Goday, MD, and ‡‡Colleen T. Lukens, PhD

ABSTRACT

To establish a foundation for methodologically sound research on the epidemiology, assessment, and treatment of pediatric feeding disorder (PFD), a 28-member multidisciplinary panel with equal representation from medicine, nutrition, feeding skill, and psychology from seven national feeding programs convened to develop a case report form (CRF). This process relied upon recent advances in defining PFD, a review of the extant literature, expert consensus regarding best practices, and review of current patient characterization templates at participating institutions. The resultant PFD CRF involves patient characterization in four domains (ie, medical, nutrition, feeding skill, and psychosocial) and identifies the primary features of a feeding disorder based on PFD diagnostic criteria. A corresponding protocol provides guidance for completing the assessment process across the four domains. The PFD CRF promotes a standard procedure to support patient characterization, enhance methodological rigor, and provide a useful clinical tool for providers and researchers working with these disorders.

Key Words: avoidant, feeding, multidisciplinary, nutrition, restrictive food intake disorder

(*JPGN* 2022;75: 351–355)

Pediatric feeding problems are heterogenous, complex, and exceedingly common (1,2). Factors in four primary domains (medical, nutritional, feeding skill, and psychosocial) contribute to the emergence and maintenance of feeding problems in pediatric populations (1). This multidomain conceptualization makes multidisciplinary research essential to elucidate the cause, consequences, and treatment of feeding concerns in pediatric populations (3,4). However, progress in this area is hindered by the absence of a common language despite a unified call for a shared conceptualization of feeding disorders from researchers and clinicians across disciplines (5). As operationalizing nomenclature and protocolizing data collection are essential to conducting quality child health research (4), developing a shared conceptualization and diagnostic framework for pediatric feeding problems is necessary to understand the etiology, epidemiology, and best practices to promote remediation of this complex condition (1,5–7).

Historically, feeding problems are often studied from discipline-specific perspectives (eg, physicians, dietitians, speech/language pathologists, occupational therapists, psychologists) (5). While this focused approach has resulted in advancements within individual areas of practice, it has limited multidisciplinary advancements which are needed to address the complexities of this patient population. Recent developments in diagnostic systems recognize limitations of single-discipline nomenclature and provide an opportunity to adopt a broader multidisciplinary framework to support clinical and research activities. In 2013, the DSM-5 established the diagnosis of avoidant restrictive food intake disorder (ARFID) to replace the DSM-IV diagnosis of feeding disorder of infancy or early childhood (8). As a diagnostic entity, ARFID captures a heterogeneous presentation of restrictive/avoidant eating spanning pediatric, late childhood, adolescent, and adult populations (9). However, ARFID diagnosis does not adequately account

for the medical and developmental skill related issues often associated with pediatric feeding concerns (1).

In 2019, Goday and colleagues (1) used the World Health Organization International Classification of Functioning, Disability, and Health (ICD) (10) framework to establish a conceptualization of feeding problems that moves even further beyond a single discipline framework. They created a diagnostic framework for PFD that retains the psychosocial dysfunction outlined in ARFID criteria, expands on the core nutritional complications highlighted in ARFID, and incorporates medical and developmental feeding skills dysfunction as additional etiological concerns. In the US, PFD has recently been added to the ICD, defined as a complex and heterogeneous disturbance in oral intake of nutrients associated with dysfunction in medical and nutritional status, feeding skills and safety, and/or psychosocial functioning.

To promote study of pediatric feeding concerns based on the conceptualization and framework provided by PFD, a method of data collection which operationalizes terms and which provides a protocol for data collection is needed (4,5). A case report form (CRF) is a specialized document used to support clinical research that is protocol driven, comprehensive in content, and standardized to permit uniform data collection (3). The primary objective in designing a CRF is to gather complete and accurate data while concurrently avoiding duplication and facilitating transcription of data from source documents onto the CRF (paper or electronic form). Development of a CRF for a specific study or content area represents a necessary pre-requisite to preserving and maintaining quality and integrity during data collection within and across institutions (3). This article describes the process of convening a panel of experts, outlines considerations for developing a patient characterization tool for the population of individuals with PFD and presents the resultant CRF.

METHODS

Expert consensus is a recognized method for establishing guidelines and theoretical frameworks related to a particular condition (11,12). The development of the PFD CRF, intended for use with children with PFD birth to 21 years, followed a mixed-method qualitative approach involving an expert panel across two phases. Phase one began with identifying experts from multidisciplinary institutions within the United States. Participation required each program involve a team of providers representing the four primary domains of PFD. In addition to multidisciplinary care teams, inclusion criteria required invited programs to deliver care through intensive inpatient and/or day hospital admissions, be affiliated with a hospital system and/or academic center, and show longevity (at least 5 years) in providing care. Inclusion criteria were intended to capitalize on program maturity and expertise and to promote uniformity in terms of practice setting and approach. (See Figure, Supplemental Digital Content, *http://links.lww.com/MPG/C853*, Inclusion Criteria Guiding Selection of PFD Expert Panel).

During a series of meetings in 2017 and 2018, workgroup members collectively identified detailed characterization of patient populations with PFD and standardization of data collection as the necessary first step in promoting increased methodological rigor in this field (11). (See Table, Supplemental Digital Content 2, *http://links.lww.com/MPG/C853*, Roster of Expert PFD Panel by Pillar and Affiliation). Focus group methodology employed with participating members involved working to achieve consensus within and across domains regarding the etiology and diagnostic features of PFD. The group collectively reviewed intake methodology employed at each of the participating sites and compiled common data elements across sites, as well as unique data points for consideration in the CRF. Potential items for a patient characterization tool were generated based on a thorough review of the literature,

expert consensus on best practices, and discussion regarding current standard of care at participating institutions. The expert panel worked collectively and in subcommittees (organized by domain) to determine the items most relevant for inclusion in the CRF.

The second phase of CRF development began in November 2020 and involved the creation of a subgroup to finalize the items to be included in the CRF (11). The members followed best practices for CRF design that support complete and accurate data collection while capturing clinically relevant information as it pertains to PFD as part of coordinated multisite research (3). As such, the development process focused exclusively on use of the CRF with a pediatric feeding disorder population. Other feeding (eg, PICA) and eating disorders (eg, anorexia nervosa) were deemed outside the scope of the CRF development process. The members streamlined the data collected at the focus group meetings and incorporated PFD diagnostic criteria (10). Through an iterative process, subcommittee members reduced items by eliminating duplicative content. Variables were further operationally defined to promote valid and reliable future data collection (11). Further revision of the CRF document occurred following multidisciplinary team review at Children's Healthcare of Atlanta, Children's Hospital of Wisconsin, and Children's Hospital of Philadelphia. The remaining four participating program reviewed and approved the resultant PFD CRF for broader dissemination.

RESULTS

The resultant PFD CRF is intended for pediatric patients experiencing feeding concerns and involves two components: (1) a patient characterization tool by domain, and (2) an instruction protocol to support valid and reliable data collection. The data collection tool contains items reflective of PFD diagnostic criteria and associated features of feeding disorders across the four domains (medical, nutritional, feeding skill, psychosocial). The user can endorse the occurrence or non-occurrence of a particular characteristic. As well, the CRF allows the user to indicate that certain information was omitted, unknown, or not collected as part of routine clinical screening (ie, not recorded – NR). The instructional protocol provides item-by-item descriptions and operational definitions to support reliable data collection in each domain and subdomain (3). Table 1 provides a summary of content by domain and links to the CRF patient characterization tool and corresponding protocols. (See Figures 3–10, Supplemental Digital Content, *http://links.lww.com/MPG/C853*, CRF and Protocols for Medical, Nutrition, Feeding Skill, and Psychosocial).

DISCUSSION

Use of methodologically sound research is necessary to enhance our understanding of PFD (6,7). Use of a CRF framework is a recognized method for promoting enhanced methodological rigor in clinical research to gather complete and accurate data about a patient population (3). Recent advances in PFD diagnostic nomenclature provided the opportunity to begin the iterative journey toward enhanced patient characterization. The goal of this work was to develop a multidisciplinary patient characterization tool to guide research on assessment and treatment and ultimately promote best clinical practice. The current study incorporated expert consensus, followed established guidelines for CRF development, and used the methodologically sound CRF framework to structure the development of a PFD CRF (3,11,13).

The resultant CRF can ultimately be used to support a patient registry. Progress in medical research is often driven by large, multisite networks involving patient registries resulting in large datasets. Large datasets promote scientific and public health advances through collating and sharing of clinical research data (13). Examples of

TABLE 1. Summary PFD CRF content by domain and subdomain.

Domain	# Subdomains	Topic	Description
Medical	3	1.1: Pregnancy and Birth	Specifies pregnancy and birth data regarding gestational age, whether the child required support in a neonatal intensive care unit (NICU), and if intubation was used to support breathing
		1.2: Medical Diagnoses	Includes conditions that potentially prohibit safe consumption of food (eg, aspiration) or delay or subvert the acquisition of feeding skill (eg, congenital heart disease)
		1.3: Diagnostic Procedures	Identifies diagnostic procedures and tests commonly employed to determine swallow safety (eg, swallow study), underlying pathology (eg, EGD) or organic conditions (eg, MRI) that may impact feeding

See Figures, Supplemental Digital Content 3: Medical Domain Case Report Form and Supplemental Digital Content 4: Medical Domain Case Report Form Protocol

Nutritional	8*	2.1: Primary Source of Nutrition	Focuses on the patient's current source(s) of nutrition, including tube feeding, drinking formula, and/or oral food consumption
		2.2: Use of Supplementation	Specifies whether the patient requires supplementation (eg, feeding tube; oral formula) and the percent of daily needs associated with supplementation
		2.3: Evidence of Dependence on Supplementation	Involves further scrutiny regarding whether patient is dependent on the supplementation to promote growth, address nutritional needs and/or support hydration, as well as source of supplementation
		2.4: Dietary diversity	Focuses on whether the diet involves items from all food groups, as well as provides an estimate of the variety within each group
		2.5: Evidence of Nutritional Deficiency	Identifies whether the patient shows evidence of nutritional deficiency and the source of the evidence (ie, laboratory analysis; diet recall)
		2.6: Dietary Adequacy by Nutrient	Specifies dietary adequacy by nutrient relative to dietary reference intake (DRI), with a recommended risk of nutrient inadequacy set at < 80% of DRI
		2.7: Anthropometric Parameters	Focuses on weight, height, and body-mass-index (BMI) with further scrutiny regarding potential signs of malnutrition based on established guidelines[13]
		2.8: Growth Curve Analysis	When two or more data points are available, assessment of potential malnutrition can involve growth curve analysis, including weight gain velocity decline, significant weight loss (ie, > 5% over 3 months), and deceleration in weight for length/height

See Figures, Supplemental Digital Content 5: Nutrition Domain Case Report Form and Supplemental Digital Content 6: Nutrition Domain Case Report Form Protocol

Feeding Skill	8	3.1 Past Therapeutic Support	Focuses on information about past therapeutic support and identifies the provider type (e.g. speech-language pathologist) and estimated duration of previous treatment
		3.2 Meal Duration	Identifies the average duration of meals (in minutes) as estimated by the caregiver
		3.3 Age Typical Diet	Involves a clinician estimate regarding whether the child is consuming an age typical diet in terms of food texture and liquid viscosity
		3.4 Current Food Texture	Focuses on whether the patient's diet involves foods that require mastication, if texture modification is required due to potential neuromuscular concerns, and the type of modification employed during meals (e.g. fork mashed; puree)
		3.5 Modified Feeding Support	Identifies modified feeding support in terms of positioning and/or special equipment utilized to permit safe or efficient eating, such as use of therapeutic seating, altered feeding position, and/or added support for stability
		3.6 Modified Feeding Strategy	Collects information about possible modifications in the delivery of food and/or liquid, including altered bite placement and use of a therapeutic utensil(s) or special bottle
		3.7 Self-Feeding	Assesses whether the child consistently and routinely engages in self-feeding during meals, with further specificity regarding the method of self-feeding (eg, finger feeding, utensil use)
		3.8 Drinking Liquids	Involves a summary of drinking format, whether thickening is required for safety concerns, and if the child demonstrates independence with depositing liquid

(Continued)

TABLE 1. *Continued*

Domain	# Subdomains	Topic	Description
Figure, Supplemental Digital Content 7: Feeding Skill Domain Case Report Form and Supplemental Digital Content 8: Feeding Skill Domain Case Report Form Protocol			
Psychosocial	5	4.1 Child Avoidance Behaviors	Focuses on barriers to achieving a nutritionally complete diet, including both active and/or passive food refusal
		4.2 Ineffective Caregiver Management Strategies	Assesses caregiver strategies aimed at improving a child's mealtime behavior, which is referred to as ineffective due to lack of resolution of the feeding dysfunction
		4.3 Disruptions in Social Functioning	Identifies the impact of feeding dysfunction on the patient's social functioning, including lack of participation in family meals, limiting participation in social events, and/or restriction in eating location
		4.4 Disruption in Parent-Child Relationship	Determines whether and how the feeding dysfunction affects the caregiver-child relationship, including poor mealtime interactions, disengagement by the child or caregiver from the meal, and caregiver stress associated with presenting foods or conducting meals
		4.5 Behavioral/Developmental Complexity	Items serve as a proxy for a child's behavioral and/or developmental functioning (as reflected by a history of receiving therapeutic and/or educational support), as well as assesses caregiver concern about the presence of problem behavior outside of meals
Supplemental Digital Content 9: Psychosocial Domain Case Report Form and Supplemental Digital Content 10: Psychosocial Domain Case Report Form Protocol			

* Subdomains span two broad nutrition topics: current pattern of dietary intake and anthropometric parameters

successful networks/patient registries include the Cystic Fibrosis Foundation (14) and Improve Care Now (15). Data from the Cystic Fibrosis Foundation's patient registry, for example, laid the groundwork for changes in nutritional care leading to improved growth and survival rates for patients based on analyzing variation in clinical care and outcomes across CF clinics in the United States and Canada (14). Similarly, the Improved Care Now network has generated substantial data working in an interdisciplinary manner, including families of patients, to guide best practices (15). A coordinated research thrust likely holds similar benefits for improving outcomes for patients with PFD; a process that starts with a uniform data collection framework to assess patients, gauge progress, and evaluate different treatment approaches within and across clinics (1,6).

The need for such a network for PFD is clear. Impaired intake of food leads to poor outcomes (1,6–8). These include impaired growth, nutritional deficiencies, dependence on enteral or oral liquid nutrition, and/or impaired psychosocial functioning. Food restriction is also associated with high levels of caregiver stress, psychiatric comorbidities, child social difficulties, and impaired family functioning (16–19). Emerging evidence suggest PFD is common and may exceed other common childhood conditions (eg, autism spectrum disorder), particularly among children with comorbid medical and/or developmental conditions (2). Estimates suggest annual prevalence of PFD between 1 in 3 and 1 in 5 among children under 5 years of age with other chronic diseases (eg, congenital or acquired respiratory, cardiac, and gastrointestinal problems) (2). High prevalence suggests potential for establishing a patient registry may hold benefit for both patients and society.

The PFD CRF may also promote best clinical practices in community settings. Use of the PFD CRF will encourage providers to adopt a multidisciplinary framework that considers the complexity of this disorder. Ideally, it also encourages providers to further assess etiological and maintaining factors potentially stemming from medical, feeding skill, and behavioral domains in addition to nutrition sequela which typically trigger attention in pediatric settings. This in turn should help providers expand clinical attention to root etiologies, as well as to collaborate with necessary multidisciplinary providers for further assessment and treatment. Ideally, changes to clinical screening based on the PDF CRF will result in improved detection of other medical concerns, patient feeding skill, and behavioral presentations (18–20).

Limitations to this work included heavy reliance on expert opinion given the absence of gold standard diagnostic PFD instruments for use in pediatric populations. Further, PFD is a relatively new diagnosis that requires further scrutiny regarding etiology and likely heterogeneity and subgroups within the diagnosis. In addition, the programs involved in this study serve children who require assessment and oversight from a multidisciplinary feeding team. This population of patients likely represents children with the most severe forms of PFD. As a result, our methodology may have overlooked aspects of patient characterization outside of patients accessing care at participating institutions and/or pediatric cohorts documented in the extant literature. Future work in this area should also include representation and involvement of the target population (ie, patients and families) to refine and improve the clinical and research utility of the PFD CRF. Moving forward, it will also be important to included provider teams outside the US serving children with complex feeding concerns.

With this work completed, future directions include field testing at participating institutions and creation of a shared patient registry/database to facilitate research in this area. Consistent with accurate data collection, this process will require establishing methods to promote reliable data collection within and across sites. Although intended to support patient characterization, creation of the PFD CRF also provides an opportunity to explore the psychometric properties of this measurement approach. The first step will involve determining feasibility of data collection across sites, including ease

of interpretation, completeness of data collection, and generalizability to the broader provider teams at participating sites. This process will guide future iterations of this PFD CRF framework.

Dissemination of this work to the broader pediatric community also holds potential to improve patient screening and promote early detection, as well as spur new lines of research inquiry associated with atypical feeding and eating. By creating a standard procedure to support patient characterization, our work is intended to serve as an initial focal point for the clinical and research community to begin to harmonize and synthesize datasets while also providing a clinical tool ready for dissemination into routine pediatric practice. As a first step, the PFD CRF is likely to undergo an iterative improvement process as our knowledge regarding the assessment and treatment of PFD expands through a more unified line of research, with an end goal of optimizing patient outcomes.

REFERENCES

1. Goday PS, Huh SY, Silverman AH, et al. Pediatric feeding disorder: consensus definition and conceptual framework. *J Pediatr Gastroenterol Nutr* 2019;68:124–9.
2. Kovacic K, Rein LE, Szabo A, et al. Pediatric feeding disorder: a nationwide prevalence study. *J Pediatr* 2021;228:126–131.e3.
3. Bellary S, Krishnankutty B, Latha MS. Basics of case report form designing in clinical research. *Perspect Clin Res* 2014;5:159–66.
4. Kahn MG, Bailey LC, Forrest CB, et al. Building a common pediatric research terminology for accelerating child health research. *Pediatrics* 2014;133:516–25.
5. Estrem HH, Pados BF, Park J, et al. Feeding problems in infancy and early childhood: evolutionary concept analysis. *J Adv Nurs* 2017;73:56–70.
6. Sharp WG, Volkert VM, Scahill L, et al. A systematic review and meta-analysis of intensive multidisciplinary intervention for pediatric feeding disorders: how standard is the standard of care? *J Pediatr* 2017;181:116–124.e4.
7. Lukens CT, Silverman AH. Systematic review of psychological interventions for pediatric feeding problems. *J Pediatr Psychol* 2014;39:903–17.
8. American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders*. 5th ed. Washington, DC: American Psychiatric Publishing; 2013.
9. Sharp WG, Stubbs KH. Avoidant/restrictive food intake disorder: a diagnosis at the intersection of feeding and eating disorders necessitating subtype differentiation. *Int J Eat Disord* 2019;52:398–401.
10. Khoury B, iKogan C, Daouk S. International classification of diseases 11th edition (ICD-11). In: Zeigler-Hill V, Shackelford TK, eds. *Encyclopedia of Personality and Individual Differences*. Cham: Springer International Publishing; 2017:1–6.
11. Holmbeck GN, Devine KA. Editorial: an author's checklist for measure development and validation manuscripts. *J Pediatr Psychol* 2009;34:691–6.
12. Lacy BE, Patel NK. Rome criteria and a diagnostic approach to irritable bowel syndrome. *J Clin Med* 2017;6:99.
13. Institute of Medicine (US). *Sharing Clinical Research Data: Workshop Summary*. Washington, DC: National Academies Press; 2013.
14. Dasenbrook EC, Sawicki GS. Cystic fibrosis patient registries: a valuable source for clinical research. *J Cyst Fibros* 2018;17:433–40.
15. Keck C, Hartley DM, Havens M, et al. Getting what is needed, when it's needed: sharing information, knowledge, and know-how in a Collaborative Learning Health System. *Learn Health Syst* 2021;5:e10268.
16. Silverman AH, Erato G, Goday P. The relationship between chronic paediatric feeding disorders and caregiver stress. *J Child Health Care* 2020;25:69–80.
17. Didehbani N, Kelly K, Austin L, et al. Role of parental stress on pediatric feeding disorders. *Child Health Care* 2011;40:85–100.
18. Greer AJ, Gulotta CS, Masler EA, et al. Caregiver stress and outcomes of children with pediatric feeding disorders treated in an intensive interdisciplinary program. *J Pediatr Psychol* 2008;33:612–20.
19. Field D, Garland M, Williams K. Correlates of specific childhood feeding problems. *J Paediatr Child Health* 2003;39:299–304.
20. Volkert VM, Sharp WG, Clark MC, et al. Modified-bolus placement as a therapeutic tool in the treatment of pediatric feeding disorders: analysis from a retrospective chart review. *J Speech Lang Hear Res* 2019;62:3123–34.