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Feeding Dysfunction in Children With Eosinophilic Gastrointestinal Diseases

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KEY WORDS

pediatric, eosinophils, feeding and eating disorders of childhood, esophagitis, child nutrition disorders, dysphagia, fibrosis

ABBREVIATIONS

GI—gastrointestinal
EGID—eosinophilic gastrointestinal disease
FD—feeding dysfunction
EoE—eosinophilic esophagitis
HPF—high-power field

Drs Furuta and Atkins contributed equally to this manuscript.

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WHAT'S KNOWN ON THIS SUBJECT: Although FD is encountered in many pediatric patients with EGIDs, specific descriptions are limited. In 1 study, 66% of patients had concomitant neurologic disease or developmental disorders; 73% of patients with FD characterized by poor oral intake responded to treatment.



WHAT THIS STUDY ADDS: Feeding difficulties are prevalent in children with EGIDs. Features of FD include maladaptive learned behaviors with altered mealtime dynamics and physical difficulties in eating mechanics. FD can persist despite effective medical treatment, affecting development of oral feeding and growth and development.

abstract



OBJECTIVES: Feeding dysfunction (FD) seen in younger children with eosinophilic gastrointestinal disease (EGID) has not been well described. Thus, our aim was to further characterize FD in children with EGIDs.

METHODS: A retrospective medical record analysis of 200 children seen over 12 months in a multidisciplinary Gastrointestinal Eosinophilic Diseases Program was performed. The clinical data of 33 children identified as also having FD were examined, including information obtained by history, physical examination, feeding evaluation, review of nutritional data, allergy testing and histologic assessment of mucosal biopsies.

RESULTS: Of 200 children with EGIDs, 16.5% had significant FD. The median age of this group was 34 months (range: 14–113 months). A variety of learned maladaptive feeding behaviors were reported in 93.9%. Frequent gagging or vomiting occurred in 84.8%. Food sensitivity was documented in 88% while 52% had other allergic disease. Twenty one percent were diagnosed with failure to thrive and 69.7% required individual or group feeding therapy. Forty-two percent had residual eosinophilia of >15 per HPF on esophageal biopsies performed at the time of symptoms.

CONCLUSIONS: FD is prevalent in children with EGIDs often presenting as maladaptive learned feeding behaviors with altered mealtime dynamics and physical difficulties in eating mechanics. FD can persist even after eosinophilic inflammation is successfully treated. Awareness of the increased prevalence of FD in children with EGIDs with enable earlier recognition of this problem, resulting in a comprehensive, individualized treatment plan with the desired outcome of improving the development, feeding, and nutrition of these children. *Pediatrics* 2010;126:e000

Eosinophilic gastrointestinal (GI) diseases (EGIDs) are a heterogeneous group of diseases characterized by idiopathic eosinophilic inflammation of the GI tract accompanied by GI symptoms.¹ In children, symptoms are often vague, leading to a delay in diagnosis of 3 to 4 years in some situations.² One often-mentioned complaint is feeding dysfunction (FD), a symptom complex typically associated with neurologic diseases, developmental delays, and occasionally gastroesophageal reflux disease.^{3–5} Although FD is commonly encountered in these conditions, specific descriptions of FD in EGIDs are limited. One series of 15 children with eosinophilic esophagitis (EoE) described FD as food refusal, oral aversion, vomiting, and poor weight gain. Improvement in these areas occurred in 73% of patients after treatment. In that series, 67% also had concomitant neurologic or developmental disease, which may have had a negative effect on feeding.⁶

No universally accepted feeding assessment protocols exist, but a number of commonly accepted feeding behavior and skill acquisition parameters are used to profile children with FD.^{7–12} Descriptive classification systems use criteria such as volume and type of food ingested, meal characteristics (eg, prolonged mealtime), and child behavior (eg, food refusal). Early identification of FD can lead to earlier treatment and positively affect child development and family quality of life.

The aim of our study was to define further FD in children with EGIDs. We took advantage of our multidisciplinary program of pediatric gastroenterologists, allergists, nurses, feeding specialists, dietitians, and psychosocial clinicians to identify and characterize FD in a well-defined cohort of children with EGIDs. FD occurred in 33 (16.5%) of 200 children who had EGIDs and did

not have comorbid diseases that typically are associated with FD. FD was characterized by a predominance of maladaptive learned behaviors (93.9%) associated with feeding and mealtimes, accompanied by a constellation of physical difficulties involving the oral motor and swallowing aspects of eating. Increased awareness and prompt symptom recognition will result in earlier diagnosis and intervention for FD.

METHODS

Study Design and Participants

We performed a single-center, retrospective analysis of medical records from 200 children who were seen in the multidisciplinary Gastrointestinal Eosinophilic Diseases Program at the Children's Hospital and National Jewish Health during a 12-month period (January 2008 to December 2008). Of these children, 33 were identified as having an EGID and FD as determined by the feeding therapists using our feeding assessment as described in the next section. Patients with other obvious causes for FD, including severe cardiac or neurologic disease, were excluded (8 patients). All available clinical data, including history and physical examination, laboratory results, imaging and endoscopy reports, family history, and allergy test-

ing, were examined in this study, which was approved by the Colorado Multiple Institutional Review Board (COMIRB 08-1208).

Feeding Evaluation

Before a clinic visit, children were identified for feeding assessment via review of available records and a detailed feeding history (Ms Haas and Ms Creskoff Maune). All children with FD were assessed during an initial or follow-up clinic visit by using a protocol developed by the Children's Hospital Feeding and Swallowing Program. This protocol addresses symptoms; observation of functional skills; learned behaviors; mealtime dynamics with caregivers; and supporting developmental skills, including gross motor, fine motor, and language skills. Results from this assessment, records from parent interviews, and direct mealtime observations by the feeding therapists formed the database for this study, which is published as supplemental information at Supplemental Table 4. Data were collected on 6 primary areas of FD, summarized in Table 1.

Nutritional Review

Available diet and growth data, including dietary recall, height, and weight, were reviewed by a pediatric dietitian

TABLE 1 Summary of FD Used in This Study

Characteristics	Definition
Learned maladaptive behaviors	Food refusal, low volume/variety of intake, poor acceptance of new foods, spitting food out, grazing, lack of mealtime structure, requiring prompting to eat, inconsistent patterns of eating
Developmental differences	Delay in gross motor, fine motor, or speech-language skills; global sensory processing dysfunction
Immature diet selection	Self-selection of purees or soft diet despite skills adequate for higher textures, preference for liquids over solids, prolonged feeding time as a result of holding food in mouth before swallowing, excessive chewing or immature oral motor skills secondary to prolonged food refusal and/or diet restrictions
Dysphagia	Swallow dysfunction manifested as food impaction or the sensation of food getting stuck in the esophagus
Oral sensory skills deficits	Altered sensory response to foods (taste, visual presentation, feel, or smell)
Oral motor skill deficits	Dysfunctional oral motor pattern for eating secondary to altered muscle tone, strength, or coordination

(Ms Henry) and a pediatrician (Dr Gilman). Nutritional parameters including BMI and height and weight z scores were calculated.

Allergy Evaluation

Results of allergy testing, including skin testing with commercial and freshly prepared extracts and food-specific immunoglobulin E levels determined by ImmunoCAP, were reviewed and collated.

Histologic Assessment

Mucosal biopsies were reviewed by pediatric pathologists (Drs Capocelli and Lovell). Examination characterized eosinophilic inflammation, reactive epithelial changes, and evidence of fibrosis as previously described.¹³ For a diagnosis of EoE, esophageal biopsies were required to have at least 15 eosinophils per high-power field (HPF) after either pretreatment with 2 months of high-dose proton pump inhibitor therapy or a normal pH or impedance study, as described previously.¹⁴

RESULTS

Clinical Characteristics of Patient Group

Clinical characteristics of children with FD are summarized in Table 2. The median age was 34 months (range: 14–113 months), with a male predominance (90.9%). Food sensitivity was noted in 88% by skin-prick or food-specific ImmunoCAP; 52% showed clinical evidence of other allergic disease. Of the 33 patients, 25 (76%) had EoE; the remainder had other EGIDs.

Upper GI endoscopy was performed on 30 of the 33 children at the Children's Hospital. The remaining 3 patients had recently undergone endoscopic examination at another institution, and the endoscopic reports were not available for review. Gross abnormalities of the esophageal mucosa included linear ridging (13 [43%] patients of 30 endos-

TABLE 2 Clinical Characteristics of Study Patients

Characteristic	Summary Description
Age, median (range), mo	34 (14 to 113)
Male gender, %	90.9
Nutritional status, median (range), z score for age	
BMI	−0.486 (−2.660 to 1.750)
Height	−0.590 (−3.900 to 1.210)
Endoscopic findings (30 patients with endoscopy reports), %	
Abnormal with linear ridging, nodularity, and exudates	47.0
Pathology findings, %	
>15 eosinophils per HPF	42.0
Fibrosis	70.0
Fibrosis in children with abnormal endoscopy	92.9
Fibrosis in children with normal endoscopy	43.8
Food-allergen sensitization, %	88.0
Sensitization on the basis of positive skin-prick test or presence of serum food-specific IgE	
Other medical issues, %	
Failure to thrive	21.0
Eczema, allergic rhinitis, or asthma	52.0
Depression or anxiety	9.0
Family medical history, %	
Dysphagia, food impaction, esophageal dilation, or stricture	18.2
Reported food or environmental allergies	55.0
EoE	12.0

IgE indicates immunoglobulin E.

copies done at our institution) and nodularity or exudates (7 [23%] of 30 patients). Twelve percent of children were noted to have a first-degree relative with EoE. An additional 18% of patients reported family members with a history of food impaction, esophageal stricture, or esophageal dilation, suggesting that some might have also had EoE. Fifty-five percent reported family members with food allergies, although no allergy testing data were available for family members.

Nutritional Analysis

A diagnosis of failure to thrive was made for 7 (21%) children. Analysis of diet records and growth data demonstrated an average z score for BMI of −0.50 and for height of −0.65 (Table 2).

Analysis of FD

Learned maladaptive feeding behaviors were the predominant form of FD reported for 93.9% of children and encompassed a range of behaviors (Fig 1, Tables 1 and 3). Gagging or vomiting was the second most common FD in these children, occurring in 84.8%.

Histologic Analysis

Esophageal biopsies performed at the time of reported symptoms of FD revealed that 42% had residual eosinophilia of >15 per HPF (15–125 eosinophils per HPF; Fig 2). Seventy percent of biopsies revealed subepithelial fibrosis. Of children with abnormal endoscopic appearance, 93% showed histologic evidence of fibrosis compared with 43.8% fibrosis in those with grossly normal mucosa (Table 2).

DISCUSSION

Frequently cited symptoms of EGIDs are vomiting, pain, dysphagia, and feeding difficulties.^{2,6,11,12,14} The objective of this study was to define the feeding characteristics of children with FD and EGIDs. Our results indicate that feeding difficulties are prevalent in children with EGIDs, with several distinct features identified, including maladaptive learned feeding behaviors with altered mealtime dynamics and physical difficulties in eating mechanics.

Maladaptive learned feeding behaviors occurred in 93.9% of our EGID co-

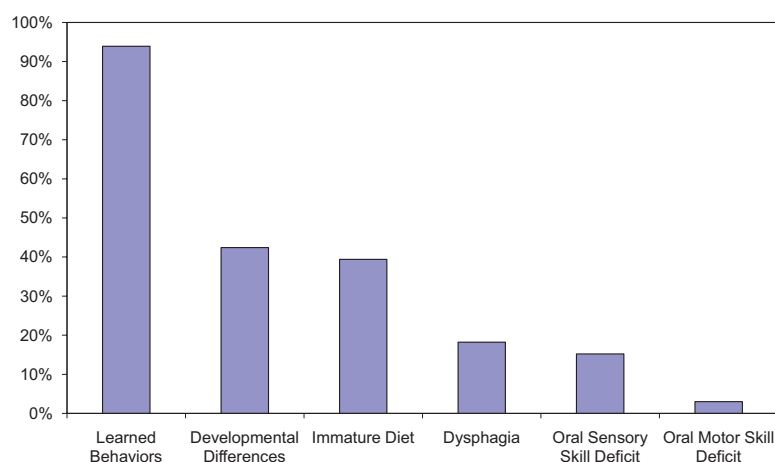


FIGURE 1

Learned feeding behaviors are the predominant form of FD seen in pediatric patients with EGID.

TABLE 3 Patients Demonstrated a Variety of Learned FDs

Learned FD	% of Patients
Low variety intake	90.9
Food refusal	87.9
Requires prompting to eat	87.9
Poor acceptance of new foods	84.8
Low volume of intake	81.8
Unstructured mealtimes	81.8
Inconsistent patterns of eating	78.8
Grazing	78.8
Easily distracted from eating	60.6
Prolonged feeding times	57.6
Holding food in mouth	27.3
Spitting food	27.3

hort with FD. This symptom complex is associated with eating, in conjunction with or followed by repetitive, unpredictable bouts of pain, which condition a child to avoid eating. These maladaptive feeding behaviors are frustrating, confusing, and unsatisfying to caregivers and can often lead to altered feeding practices in an effort to provide the child adequate nutrition.^{7,15,16} In an effort to minimize pain or discomfort, children with EGIDs may reduce volume or variety of foods, and caregivers

may inadvertently reinforce undesired behaviors. For example, when a child refuses offered foods and is eventually provided preferred foods, a pattern that unintentionally reinforces food refusal develops. Although well intentioned, this response establishes a cycle of behavior and reinforcement that results in greater rigidity and maladaptive feeding behavior, leading to stressful mealtime dynamics. Consistent with previous reports, we found that gagging and vomiting are frequent symptoms of EGIDs in children, occurring in 84.8% of our patients.^{12,15} In addition to being an identified symptom of EGIDs, gagging and vomiting may occur secondary to altered oral motor and/or oral sensory skills or as a learned response to avoid eating. The importance of learned behaviors in these patients is highlighted by the persistence of FD in the 58% of patients who had resolution of esophageal inflammation on their most recent biopsy.

Swallowing dysfunction is frequently seen in older children and adults with esophageal disease as dysphagia manifesting as food impaction or the sensation of food “getting stuck.”¹² In our patient population, solid food avoidance and preferential liquid and puree selection were believed to be manifestations of esophageal swallowing problems in the younger age group. The exact pathophysiology remains unknown, but it is possible that this diet preference stems from persistent inflammation, as suggested by the finding that approximately half of the children had mucosal eosinophilia. Alternatively, this symptom could result from tissue remodeling without inflammation, as suggested by gross endoscopic features of ridging and histologic evidence of fibrosis. In young children who have not yet developed adequate expressive language

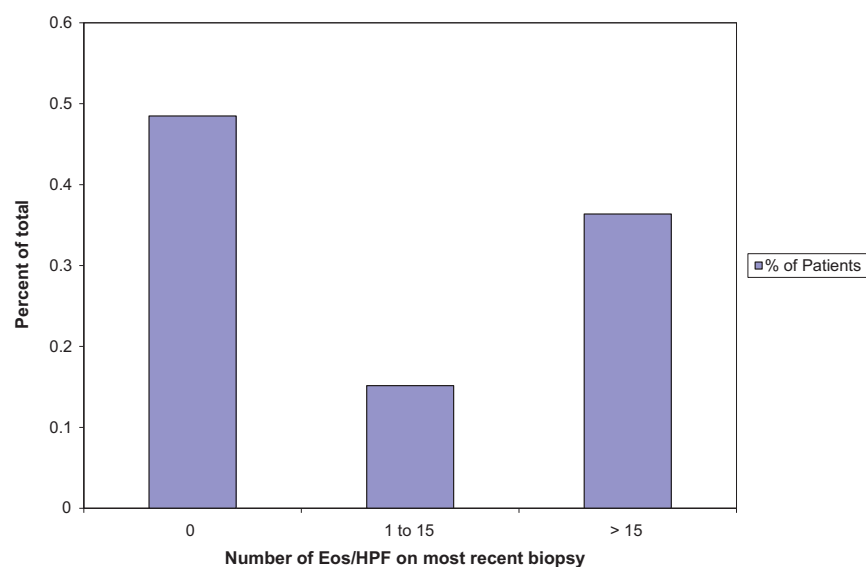


FIGURE 2

FD can persist after eosinophilic inflammation has resolved.

skills, caregivers may miss these symptoms.

Identification of FD early in the course of children's disease is important for several reasons. First, our data suggest that EGIDs can have a negative impact on oral motor skill development for age-appropriate diet advancement. Medical intervention may involve liquid elemental diets or significant restriction of the range of foods or textures that can be offered to children, resulting in a lack of developmentally appropriate learning opportunities with more advanced foods. In fact, children tended to prefer an immature diet rather than display true oral motor skill deficits (see definitions, which are published as supplemental information at Supplemental Table 4). Second, FD may contribute to deficient oral intake, leading to malnutrition and growth disturbance, as seen in 21% of our patients. Third, children with EGIDs often require consultation with an experienced pediatric feeding specialist once inflammation is treated and the appropriate diet is identified. Either individual or group therapy was recommended for 23 (69.7%) children in our series to address learned maladaptive feeding behaviors, oral motor immaturity, volume of intake, and variety of intake. When appropriate, children were seen for group therapy to cap-

italize on peer influence and provide an opportunity for parents to support one another. In all cases, families were given a home program to promote appropriate feeding skill development via strategies that could be implemented readily in the home environment. Many of these strategies, including feeding children on a routine, keeping a predictable daytime schedule, and having children sit consistently at the table, are helpful for all children, even those who are fed elemental formula exclusively.

One limitation of this study is its retrospective nature. Although the number of children included is relatively small, this represents the largest group of children who had EGIDs and in whom FD has been carefully examined. Characterization of FD was performed with a nonvalidated protocol, because no other tools exist to date. There also may be some referral bias in that, as a tertiary referral program, we may evaluate subsets of patients who have more severe symptoms as a result of advanced disease. This may be reflected in the relatively high rate of fibrosis seen in our patient population, although it is notable that several studies also commented on increased subepithelial fibrosis seen in EoE.^{13,17}

Eating is a dynamic learning experience. EGIDs can derail the development of oral feeding and alter family mealtime dynamics. Despite effective medical treatment, the impact on feeding can persist and must be addressed. Although several symptoms are similar in children with EGIDs and those with gastroesophageal reflux disease, in this study, we identified several distinct characteristics of FD that are unique to the young child with EGIDs. These findings should alert general pediatricians, pediatric gastroenterologists, and allergists that FD is a prevalent and pervasive set of symptoms that adversely affect children with EGIDs. As such, it is important to ask all patients and families who are affected by EGIDs about their feeding experiences and refer them to feeding experts when appropriate. In addition, increased knowledge about the co-occurrence of EGIDs and feeding problems may prompt evaluation for EGIDs in feeding patients without a clear cause for their FD.

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