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Onset, Trajectory, and Pattern of Feeding Difficulties in Toddlers Later Diagnosed with Autism

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Abstract

Objective: To examine the emergence and trajectory of feeding difficulties in young children who are later diagnosed with autism spectrum disorder (ASD).

Method: The Behavioral Pediatrics Feeding Assessment Scale (BPFAS) was administered to a sample of 93 toddlers with an older sibling with ASD -- the high-risk group -- and 62 toddlers with no known familial ASD -- the low-risk group -- as part of a larger infant sibling study. The BPFAS was completed by parents at 15, 18, 24, and 36 months of age. At 36 months, participants underwent a diagnostic assessment and were classified into one of four outcome groups: ASD, Non-Typical Development, High-risk Typically Developing, and Low-risk Typically Developing. The BPFAS was scored for total frequency of feeding difficulties, as well as autism-specific factor scores previously described in the literature.

Results: The frequency of feeding difficulties increased significantly more rapidly in the ASD group between 15 and 36 months of age, and by 36 months, they exhibited a significantly higher total frequency score than all other groups. Analysis of the factor scores revealed a similar pattern for the food acceptance and mealtime behavior domains, but no significant differences in the medical/oral motor domain.

Conclusion: Feeding difficulties develop significantly more rapidly in children with ASD, with longitudinal monitoring revealing the steeper trajectory earlier than can be detected with cross-sectional analysis. Children with ASD are at risk of health and social consequences of poor feeding behavior that may potentially be minimized if addressed early and appropriately.

Index Terms:

Autism; feeding difficulties; mealtime behavior; food selectivity; social communication

Introduction

One of the primary responsibilities of parents and other primary caregivers of children is to feed them. From the first hours of life, feeding becomes a daily interactive activity in which parents are highly invested. Success in this endeavor is a source of parental satisfaction and can become an enjoyable family social activity that contributes to family cohesion. Parents want their children to eat nutritious foods and gain appropriate weight. Primary healthcare providers have similar goals and diligently monitor children's growth and weight gain. While many children are enthusiastic eaters, anywhere between 6–50% of parents experience their children as “picky” eaters.^{1,2} Difficulties feeding one's child may result in concerns regarding nutrition and health as well as frustration with the child's behavior.¹ Given the frequency of picky eating, perhaps it is not surprising that pediatricians may consider it a normal developmental phenomenon, particularly when the child is growing and gaining weight normally, and not recognize that a serious problem is emerging. However, picky eating can result in disordered feeding relationships between caregivers and children. The health and developmental implications of picky eating and other behavioral feeding difficulties have not yet been studied longitudinally.²

Feeding difficulties are well recognized in children with autism spectrum disorder (ASD), who are five times more likely to have feeding problems than typically developing children.³ The DSM-5⁴ has included extreme food restriction as a manifestation of a core symptom of ASD within the category of insistence on sameness/inflexible adherence to routines. The commonly identified difficulties are restricted food preferences (e.g. food selectivity) based on sensory properties of foods (texture, taste, smell, color, packaging, presentation), fear of novel foods (neophobia - a form of food selectivity), prolonged mealtime durations, and negative mealtime behaviors associated with resistance to non-preferred foods or other mealtime expectations or routines. Cornish's descriptive study⁵ revealed that only 6% of children with ASD ate the recommended amount of fruits and vegetables, and that 53% of these children had nutritional deficiencies. Such atypical patterns of food intake are often not detected during pediatric well child visits since nutritional status is screened with anthropometric measures, and despite their unbalanced nutrient intake, most children with ASD demonstrate normal weight and growth.³

Despite the well-established prevalence of feeding difficulties in children with ASD, much has yet to be documented about etiology and course of development. Many studies have touched upon age of emergence, although the cross-sectional nature of these investigations provides only a snapshot of a more continuous process. Previous research has not yet explored how emerging feeding problems may be related to the onset of core ASD symptoms.

The objective of this study was to provide a longitudinal perspective on the development of feeding behavior, examining the evolution over time of the feeding problems that have been well-characterized in older children with ASD. In this study, we compared trajectories from 15 to 36 months of typically developing children to those who are later diagnosed with ASD and examined associations with social-communication delays and other symptoms of ASD.

Method

Sample characteristics

The sample included 93 toddlers with an older sibling with ASD -- the high-risk (HR) group -- and 62 toddlers with no known familial ASD -- the low-risk (LR) group. The sole inclusion criterion for the HR group was status as a younger sibling of a child with ASD; exclusion criteria included birth before 32 weeks of gestation and any known genetic disorder (e.g., Fragile X syndrome, Tuberous Sclerosis, etc.) in the older affected sibling or family. The primary inclusion criterion for the LR group was status as a younger sibling of a child (or children) with typical development, while exclusion criteria were birth before 36 weeks of gestation, neurodevelopmental or learning disorders in any older sibling, and ASD in first-, second-, or third-degree relatives. These children were participants of a larger longitudinal infant sibling study that monitors early development to better characterize early manifestations of ASD. All participants were first seen at either 6 or 9 months of age and assessed prospectively at regular intervals up to 36 months of age. A feeding questionnaire was completed by parents at 15, 18, 24, and 36 months. At 36 months, comprehensive diagnostic assessments were used to classify participants into one of three outcome groups based on scores from the Autism Diagnostic Observation Schedule, 2nd Edition (ADOS-2) and the Mullen Scales of Early Learning. The ASD group (n=19) met DSM-5 criteria for autism spectrum disorder and scored above the ASD cutoff on the ADOS-2. The Non-Typical Development group (Non-TD; n=24) did not meet DSM-5 criteria for ASD and had ADOS-2 scores ≥ 3 points below the ASD cutoff and/or two or more Mullen subscale scores ≥ 1.5 standard deviations (SD) below the normative mean and/or one or more Mullen subscale scores ≥ 2 SD below the mean. The Typically Developing (TD) group had ADOS-2 scores ≥ 3 points below the ASD cutoff, no more than one Mullen subscale score ≥ 1.5 SD below the mean, and no Mullen subscale score ≥ 2 SD below the mean. The TD group was further stratified by risk group (HR-TD, n=54 and LR-TD, n=58). Sample characteristics are shown in Table 1.

Measures

Behavioral Pediatrics Feeding Assessment Scale (BPFAS)⁶: The BPFAS is a 35-item caregiver-report questionnaire that assesses a variety of problematic and desirable eating behaviors using a five-point Likert scale (never to always). Previous studies have demonstrated that the BPFAS provides a reliable (e.g., Cronbach's alpha $> .80$) and valid estimate of feeding problems across a range of pediatric populations.⁶⁻⁸ The first 25 items are frequency ratings of child feeding behaviors ('Has a poor appetite;' 'Gets up from table during meal'), while the last 10 items measure the respondent's feelings about and strategies for dealing with the child's feeding behaviors ('I get frustrated or anxious when feeding my child;' 'I coax my child to get him/her to take a bite'). Each of the 35 Likert-scale items is followed by a dichotomous question querying whether the respondent considers the feeding behavior problematic. The primary variable derived from the BPFAS is the total frequency score⁶, which is a summary of the 35 Likert-scale item ratings, with desirable behaviors reverse-scored so that higher scores indicate higher frequencies of feeding difficulties. The dichotomous "problem" questions following each item are not included in the total frequency score. Additional analyses separated child-specific behaviors (items 1-25) from

respondent concerns (items 26–35). Scores were also calculated for three previously derived factors.^{6–8} The food acceptance factor includes items such as ‘Will try new food’ and ‘Eats vegetables’ to measure the degree of food refusal. The medical/oral motor factor includes items such as ‘Has problems chewing food’ and ‘Chokes or gags at mealtime’ to assess mechanical physiological barriers to feeding. The mealtime behavior factor includes items such as ‘Comes readily to mealtime’ and ‘Tantrums at mealtimes’ to quantify the degree of problematic behavior.

Autism Diagnostic Observation Schedule 2nd edition (ADOS-2)⁹: The ADOS-2 is a standardized play-based observational assessment that measures communication, social, play, and repetitive behavior characteristics. Psychometric studies report high interrater reliability and agreement in diagnostic classification (ASD versus non-ASD).⁹ Items are rated on a four-point scale ranging from developmentally appropriate to severely autistic. These items are summarized and translated into a severity score ranging from 1 to 10 (scores of 4 and above are in the ASD range). The ADOS-2 was given at 18, 24, and 36 months and used in outcome determination.

Mullen Scales of Early Learning (MSEL)¹⁰: The MSEL is a developmental test for infants and young children that is normed up to 68 months and has excellent internal consistency (median=0.91) and test-retest reliability (median=0.84).¹⁰ It assesses ability in four domains: fine motor, visual reception, expressive language, and receptive language. Raw scores are converted into age equivalent and T-scores. This test was conducted at each visit.

Imitative Sequences¹¹: A series of actions (e.g., banging table, clapping hands, wiggling tongue) were performed by the examiner, each followed by a short pause allowing the child the opportunity to imitate. Imitations were rated as perfect pass, partial pass, or fail (not attempted) and used to generate a total pass proportion. Examiners were trained to 80% reliability in scoring.

Social-communication behavioral coding: Video recordings of the MSEL and a standardized parent-child play interaction were coded for gaze behavior using a previously validated coding system.¹² Coding was done by researchers unaware of risk group or outcome classification who maintained reliability at or above 80% a master coder throughout the project. This data was then used to calculate the frequency of gaze to faces per minute as an index of social-communication competence. This and the imitation measure were used to examine how feeding problems were related to the emerging social and communication delays of ASD.

Analysis Plan

Data analysis was conducted using hierarchical generalized linear modeling (HGLM) to examine growth curves, wherein change over time in the BPFAS total frequency scores were modeled as a continuous variable for the 4 outcome groups between 15 and 36 months. Models included both level-1 random intercepts and slopes for individual subjects, and fixed effects for age, outcome group, and the interaction between age and outcome group. The

latter interaction term was used as a way to test the hypothesis that the ASD group would show greater increases in feeding difficulties over time when compared to the other 3 groups. For factor scores derived by Allen et al.⁷, the HGLM analytic models employed a Poisson distribution with a log-link in order to better fit the data, given severe right skewness. Scores were rescaled to have a minimum of zero for these analyses. Following analyses of frequency and factor scores, we then examined imitation and gaze behavior as possible mediators or moderators of outcome and age effects, hypothesizing that feeding difficulties may reflect, and be manifestations of, social-communication delays that are components of ASD. Mediation effects were tested by reexamining group and age effects in the presence of covariate-only models, compared to models without any covariates. Moderation effects were tested by examining interaction effects between covariates and group and age. All main and interaction effects of interest were tested using a traditional likelihood ratio test, wherein the numerical difference of -2LogLikelihood fit indices was tested using the difference in the models' degrees of freedom as the degrees of freedom in a chi-square test. Thus all tests are expressed using chi-square values. All analyses were conducted in R¹³, version 3.2.2, using the lme4 package.¹⁴

Results

Analysis of the BPFAS total frequency score revealed a significant main effect for age ($X^2=31.64$, $df=1$, $p<.001$), with an increase of 0.36 (.06) points on the BPFAS per month between 15 and 36 months. The main effect for group was marginally significant ($X^2=7.01$, $df=3$, $p=.07$), whereas the interaction between group and age was highly significant ($X^2=19.41$, $df=3$, $p<.001$). Further analyses of both child-specific and parent-specific frequency scores revealed a consistent pattern, both with marginally significant main effects for group and highly significant interactions between group and age. Simple comparisons between groups revealed that the ASD group exhibited a significantly increasing total frequency score between 15 and 36 months compared to all other groups, and that by 36 months, they exhibited a significantly higher total frequency score overall than all other groups (Figure 1). No differences were found between any of the other groups in change over time or at any given age point (Table 2).

The analysis of the 3 factor scores revealed a similar pattern for the food acceptance and the mealtime behavior domains, with significant age by group interaction effects ($X^2=9.40$, $df=3$, $p=0.02$ and $X^2=15.88$, $df=3$, $p=0.001$, respectively), where the ASD group exhibited greater increases in feeding difficulties over time compared to the other groups. For the medical/oral motor domain, there were no main effects for either age or group, and no significant interaction between age and group (p 's > 0.3).

Additional covariate analysis of imitation showed neither mediational effects ($X^2=0.04$, $df=1$, $p=0.84$) nor moderation effects ($X^2=2.32$, $df=3$, $p=0.50$) on age and outcome. While gaze behavior did show significant differences between outcome groups, covariate analysis of gaze behavior also showed neither mediational ($X^2=0.52$, $df=1$, $p=0.47$) nor moderation effects ($X^2=0.90$, $df=3$, $p=.83$) on age and outcome (Table 2).

Discussion

This study showed an increasing frequency of parent-reported feeding difficulties over time in all four groups, confirming the widely accepted notion that parent-perceived feeding difficulties are present and increase over time in all children during the first few years of life.¹⁵ This increase in feeding difficulty, however, developed significantly more rapidly in children with ASD compared to the other three groups, resulting in significantly more feeding difficulties by 36 months of age than any other group. Thus, while developmental trajectories of feeding difficulties escalated in this sample of children with ASD from 15 to 36 months relative to the other groups, they were not revealed by cross-sectional analyses until 36 months, highlighting the usefulness of longitudinal monitoring. Analyses of individual feeding factors demonstrated that feeding difficulties in children with ASD were not due to oral motor problems (e.g., difficulty chewing food, choking or gagging) but involved challenges in both food acceptance (e.g., trying new food, eating vegetables) and mealtime behaviors (e.g., getting up from table during mealtime, tantrums when presented with non-preferred foods). We hypothesized that feeding difficulties might be related to the social-communication deficits that are part of the emerging ASD phenotype, but the lack of significant interactions between feeding behavior and either imitation or social gaze to adults did not support this hypothesis. Alternatively, we suggest that feeding difficulties result from a convergence of factors associated with ASD including sensory sensitivities, preference for sameness, diminished responsiveness to social reward, increased reactivity in response to frustration, and possibly other as yet unidentified mechanisms.

The increase in and persistence of feeding difficulties in children with ASD over time may have a range of negative medical, developmental, and social consequences. Nutritional deficits, cognitive, cardiovascular and metabolic consequences have been described.^{3,16–18} Persistent negative mealtime behaviors can also disrupt family social dynamics. Parents of children with ASD who display resistant mealtime behaviors report disappointment in the quality of their family mealtime interactions.¹⁹ The disruptive behavior can diminish willingness to participate in family meals, resulting in separate mealtimes for different family members instead of the desired family meals that pediatric health care professionals recommend.²⁰

These health and social consequences of behavioral feeding issues can be harmful to the child and family if left unaddressed. Primary health care providers (PCPs) routinely monitor patient growth and may inquire about feeding concerns during office visits. However, children with ASD often maintain a healthy growth trajectory, and parents may not always present such concerns when pressed for time in brief well-child visits when there are other seemingly more salient developmental and behavioral issues to discuss. Given that picky eating is so common starting in the early years of life, parents and the PCP are clearly challenged to differentiate symptoms of selective eating due to ASD from picky eating in the typically developing child. Routine surveillance and screening for the presence and severity of feeding-related concerns during well-child visits may be warranted, especially in young children with red flags for ASD. In particular, PCPs may want to inquire about the range of different foods the child will eat, the extent and rate of narrowing of food preferences, whether the child insists on specific commercial brands of preferred foods or very specific

routines, how long mealtimes last (more than 30 minutes), whether distractions are required in order to feed the child (e.g., cell phone, television, specific toys), and the quality of mealtime social interactions to help decide, with the family, whether and when additional evaluation and intervention are indicated.

Given the pervasiveness, intensity, and duration of feeding problems in individuals with ASD, the need for preventative strategies and evidence-based treatments is clear. Johnson et al.²¹ recently piloted a parent training program for feeding problems in children with ASD ages 2 through 7 years of age. This randomized trial with wait list control was delivered in eleven 60–90 minute sessions over the course of 20 weeks. The program was rated as acceptable and feasible for parents to use at home. It resulted in improved parent confidence to address feeding problems in their child. Feeding and disruptive behaviors were significantly reduced and both food selectivity and mealtime behavior issues improved significantly relative to the control group.²¹ Another study of a different feeding program reported improvements in parental self-efficacy, mealtime difficulty, and range of foods consumed after implementation of an educational intervention for parents of young children with ASD.²² These and further research studies are needed to develop evidence-based guidelines and practical resource for parents and to help improve mealtime behavior issues and food selectivity before they exacerbate the health and social challenges that children with ASD already face.

The current study demonstrates that feeding problems in children with ASD appear early and escalate more quickly than in typically developing children with picky eating. Given that feeding difficulties are now considered part of the ASD diagnostic phenotype, our finding may be added to other early “red flags” for ASD that parents, pediatricians and other health care providers use. Further research to help differentiate emerging ASD patterns of food restriction from regular “picky eating” is needed to help caregivers and health care professionals identify and begin treatment as early as possible.

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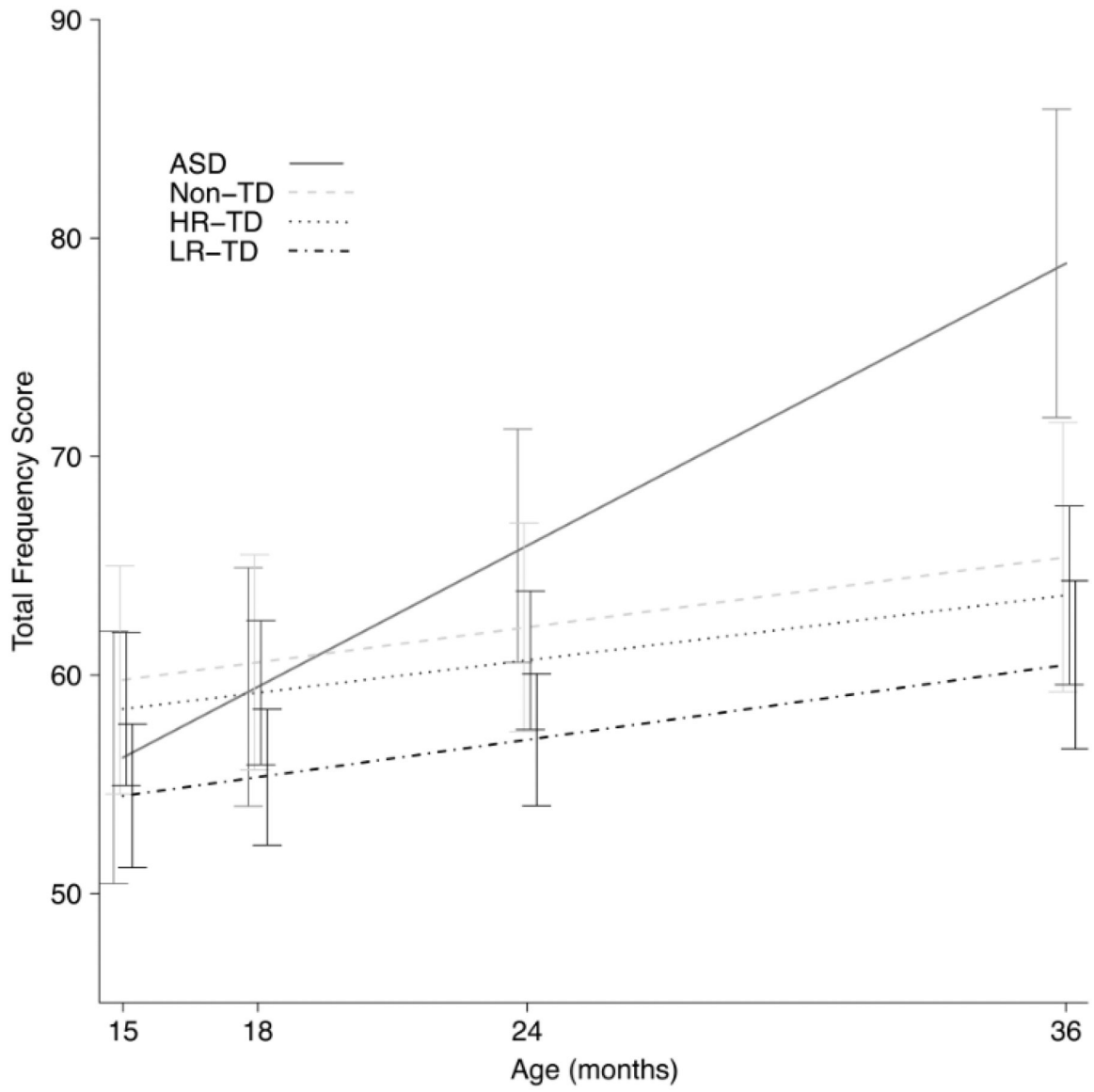


Figure 1:
Total feeding problems frequency as a function of age and outcome group.

Table 1:

Sample characteristics and outcome assessment descriptive statistics.

	ASD	Non-TD	HR-TD	LR-TD
Sample characteristics				
Sample size	19	24	54	58
Sex (% male)	79.0%	70.8%	55.6%	58.6%
Recruitment age in months	2.71 (3.6)	2.88 (3.6)	2.73 (4.3)	4.66 (2.8)
Race (% non-white)	23.5%	40.9%	38.5%	33.9%
Household income (% \$100k)	18.8%	39.1%	51.0%	38.2%
Maternal education (% college degree or higher)	64.7%	63.6%	68.0%	80.4%
Maternal age at birth in years	34.1 (5.3)	35.1 (3.9)	35.4 (4.9)	32.2 (5.5)
Paternal age at birth in years	36.6 (6.8)	39.0 (6.7)	37.4 (5.3)	34.1 (5.0)
MSEL age equivalents				
Fine motor	29.6 (7.1)	31.7 (8.6)	36.1 (4.6)	34.6 (4.1)
Visual reception	30.9 (10.9)	37.1 (11.5)	43.1 (7.0)	43.9 (6.3)
Expressive language	25.9 (8.1)	31.1 (7.4)	36.8 (4.9)	38.6 (4.4)
Receptive language	24.3 (9.4)	32.3 (7.5)	34.9 (5.3)	37.8 (5.5)
ADOS-2 severity scores				
Total severity	6.9 (1.6)	2.8 (1.3)	1.4 (0.5)	1.3 (0.4)
Social/affect severity	6.4 (1.8)	3.2 (1.7)	1.9 (0.8)	1.8 (0.7)
Repetitive behavior severity	7.9 (1.7)	4.6 (2.4)	3.1 (2.3)	2.2 (2.0)

Note: Standard deviations are in parentheses

ASD = autism spectrum disorder; Non-TD = Non-Typical Development; HR-TD = high-risk typically developing; LR-TD = low-risk typically developing; MSEL = Mullen Scales of Early Learning; ADOS-2 = Autism Diagnostic Observation Schedule, 2nd edition

Table 2:

BPFAS and covariate means and standard deviations.

	ASD	Non-TD	HR-TD	LR-TD	P-value
Total frequency score					
15 months	55.47 (17.0)	59.29 (15.9)	56.32 (13.3)	53.63 (7.6)	0.41
18 months	59.88 (17.0)	60.55 (16.1)	60.46 (14.9)	55.00 (11.0)	0.20
24 months	67.24 (13.1)	60.29 (16.3)	61.73 (13.4)	58.53 (12.4)	0.13
36 months	79.93 (10.7) ^a	65.76 (18.3) ^b	62.87 (15.4) ^b	60.23 (12.7) ^b	<0.001
Food Acceptance factor					
15 months	10.87 (5.0)	10.41 (3.5)	10.12 (3.4)	10.20 (3.2)	0.91
18 months	11.75 (4.9)	12.05 (4.9)	11.72 (4.3)	11.08 (3.5)	0.78
24 months	15.41 (4.3) ^a	10.59 (3.8) ^b	12.30 (4.0) ^b	11.36 (3.4) ^b	<0.001
36 months	17.33 (3.4) ^a	12.00 (4.5) ^b	12.30 (4.4) ^b	11.96 (3.8) ^b	<0.001
Medical/Oral Motor factor					
15 months	15.67 (5.2)	16.94 (4.5)	16.61 (5.0)	15.69 (3.1)	0.62
18 months	17.38 (5.7)	16.35 (5.0)	16.76 (4.8)	14.87 (3.8)	0.12
24 months	16.35 (4.3)	15.24 (4.8)	15.98 (4.7)	15.35 (3.8)	0.77
36 months	16.53 (2.9)	16.38 (5.0)	15.38 (4.2)	14.93 (3.1)	0.32
Mealtime Behavior factor					
15 months	14.33 (5.2)	16.59 (5.3)	15.42 (4.5)	13.86 (3.0)	0.09
18 months	15.75 (5.3)	17.90 (5.5)	17.24 (5.5)	15.52 (4.1)	0.12
24 months	19.18 (5.2)	19.29 (7.7)	18.93 (5.5)	18.20 (5.1)	0.84
36 months	25.80 (5.7) ^a	21.10 (6.6) ^b	20.06 (5.9) ^b	19.80 (5.7) ^b	0.006
Imitative Sequences (proportion passed)					
15 months	0.27 (0.21)	0.37 (0.27)	0.26 (0.18)	0.32 (0.25)	0.43
18 months	0.30 (0.19)	0.42 (0.30)	0.38 (0.26)	0.38 (0.29)	0.75
24 months	0.53 (0.34)	0.36 (0.29)	0.48 (0.35)	0.50 (0.30)	0.60
36 months	0.64 (0.37)	0.62 (0.46)	0.67 (0.31)	0.71 (0.35)	0.84
Social Gaze to Face (frequency per minute)					
15 months	2.70 (1.80) ^a	2.76 (1.80) ^a	3.54 (1.80) ^a	4.68 (2.40) ^b	<0.001
18 months	1.74 (0.60) ^a	2.28 (1.80) ^{ab}	2.94 (1.80) ^{bc}	3.36 (1.80) ^c	0.001
24 months	1.68 (1.20)	2.52 (1.80)	2.40 (1.80)	2.76 (1.20)	0.11
36 months	1.32 (0.60) ^a	3.42 (3.00) ^b	3.36 (1.80) ^b	4.02 (2.40) ^b	0.002

Note:

Standard deviations are in parentheses

p-value reported is for group main effect at each age

Groups with different superscripts differ significantly at p<.05

BPFAS = Behavioral Pediatrics Feeding Assessment Scale; ASD = autism spectrum disorder; Non-TD = Non-Typical Development; HR-TD = high-risk typically developing; LR-TD = low-risk typically developing