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Abstract

Purpose: Cluttering is a fluency disorder that has been noted clinically in individuals with fragile X syndrome (FXS). Yet, cluttering has not been systematically characterized in this population, hindering identification and intervention efforts. The present study examined the rates of cluttering in young adult males with FXS using expert clinical opinion, the alignment between expert clinical opinion and objectively quantified features of cluttering from language transcripts, and the association between cluttering and aspects of the FXS phenotype.

Method: Thirty-six males with FXS (18-26 years; *M* = 22, *SD* = 2.35) contributed language samples and completed measures of nonverbal cognition, autism symptoms, anxiety, and symptoms of attention deficit/hyperactivity disorder (ADHD). The presence of cluttering was determined by the consensus of two clinical experts in fluency disorders based on characteristics exhibited in the language sample. Cluttering features (speech rate, disfluencies, etc.) were also objectively quantified from the language transcripts.

Results: Clinical experts determined that 50% of participants met the criteria for a cluttering diagnosis. Phrase repetitions were the most salient feature that distinguished individuals who cluttered. Although the presence of cluttering was not associated with autism symptoms or mean length of utterance, cluttering was more likely to occur when nonverbal cognitive ability was higher, ADHD symptoms were elevated, and anxiety symptoms were low.

Conclusions: Half of the young adult males with FXS exhibited cluttering, which supports FXS as a genetic diagnosis that is highly enriched for risk of cluttering. Cluttering was associated with increased ADHD symptoms and cognitive ability, and reduced anxiety symptoms. This study contributes a new description of the clinical presentation of cluttering in males with FXS and

may lead to improved understanding of the potential underlying mechanisms of cluttering and eventual refinements to treatment and diagnosis.

Cluttering in the Speech of Males with Fragile X Syndrome

Cluttering is a speech-language fluency disorder characterized by reduced intelligibility due to issues related to a perceived rapid or irregular rate of speech. It is different from stuttering in that people who stutter have motoric difficulty maintaining the forward flow of speech, whereas in cluttering, individuals are perceived to produce speech at a rapid or irregular pace which reduces intelligibility or causes the message to break down (Scaler Scott & St. Louis, 2009; St. Louis & Schulte, 2011; Van Zaalen- op ’t Hof et al., 2009). Both stuttering and cluttering have been found to co-occur with neurodevelopmental disabilities such as autism spectrum disorder (ASD); however, cluttering rarely occurs in isolation (Scaler Scott et al., 2014; St. Louis & Schulte, 2011; Van Borsel & Vandermeulen, 2009). The rate of cluttering among individuals with intellectual disability appears to be substantially elevated relative to the general population, with emerging evidence suggesting that as many of 50% of individuals with intellectual disability exhibit clinically significant cluttering (Coppens-Hofman et al., 2013). Although fragile X syndrome (FXS), the leading inherited cause of intellectual disability (Rousseau et al., 1994), has often been claimed to be associated with cluttering, there have been few systematic empirical investigations of the prevalence, presentation, or correlates of cluttering within this group. The present study focused on the occurrence and presentation of cluttering within young adults with FXS and relationships between cluttering and other aspects of the FXS phenotype in an effort to inform assessment and treatment.

Characterization of Cluttering

Cluttering is a clinical disorder of speech fluency diagnosed by speech language pathologists (SLPs) related to a perceived abnormal rate and overall reduced intelligibility (Scaler Scott & St. Louis, 2009; St. Louis et al., 2003, 2007; Van Zaalen- op ’t Hof et al., 2009).

Although the prevalence of cluttering in the general population is unknown, St. Louis et al. (2010) estimate approximately 50% of people in the United States know a person who clutters. A common set of criteria used by clinicians and experts to diagnose cluttering is the Lowest Common Denominator (LCD; Scaler Scott & St. Louis, 2009; Scaler Scott, 2020; St. Louis & Schulte, 2011), which relies on a handful of perceptual features of speech. According to the LCD definition, perceived rapid or irregular speech rate for the speaker is a mandatory feature; however, the perception of rapid/irregular rate has proven difficult to quantify objectively. Studies show that the speech rate of individuals who clutter is not always objectively faster than average, despite the perception of faster rate (Bretherton-Furness & Ward, 2015; Van Zaalen- op 't Hof et al., 2009). It is posited, therefore, that the perceived rapid rate is specific to the speaker, where instances of more rapid rate within a speaking context surpass the intrinsic rate needed for articulate fluent speech (St. Louis & Schulte, 2011).

"Bursts" of more rapid speech may cause successive breakdowns that manifest as speech production errors, such as disfluencies, over-coarticulation, and atypical pauses (Bakker et al., 2011; Scaler Scott, 2020; Scaler Scott & St. Louis, 2009). Thus, in addition to perceived rapid or irregular speech rate, the LCD definition requires the presence of at least one of the following features: excessive "normal" disfluencies, excessive over-coarticulation, and/or excessive atypical pauses (St. Louis & Schulte, 2011). Disfluencies are defined as phrase repetitions, multisyllabic word repetitions, single-word and syllable repetitions without tension, revisions (i.e., "I [went] ran to the store"), and interjections (i.e., "I [uh] ran to the store"). Over-coarticulation includes an excessive degree of influence from surrounding words, causing syllable and sounds to be overly blended or omitted. Atypical pauses are defined as pauses that occur in places other than at syntactic boundaries (St. Louis & Schulte, 2011).

When diagnosing cluttering using the LCD definition, SLPs must use clinical experience to judge the impact on the speaker's intelligibility (Scaler Scott & St. Louis, 2009). There are no specific criteria for a perceived fast speech rate or what constitutes "excessive" occurrences of disfluencies and over-articulation. Thus, the LCD definition relies on perceptual judgment rather than objective data. The current study was designed to compare expert clinical opinion using the LCD definition to objective measurements of LCD characteristics such as rate of speech and relative frequency of over-articulations and disfluencies. It was hypothesized that this comparison could provide complementary objective LCD data useful for clinicians assessing cluttering.

Cluttering in Individuals with FXS

FXS is an X-linked neurodevelopmental disorder that occurs in approximately 1:7000-11,000 individuals (Hunter et al., 2014) caused by an excessive generation of repeats of the CGG trinucleotide sequence on the Fragile X Mental Retardation (*FMR1*) gene (Hunter et al., 2014). The *FMR1* gene is responsible for making fragile X mental retardation protein (FMRP) essential to cognitive development, and FMRP is reduced or absent in FXS (Hagerman et al., 2017). Males are typically more severely impacted than females, with the majority of males with FXS having an intellectual disability and about 60% meeting diagnostic criteria for ASD (Abbeduto et al., 2019; Hallin et al., 2016; Harris et al., 2008; Klusek et al., 2014). Children with FXS frequently have symptoms or co-occurring diagnoses of anxiety and/or attention-deficit hyperactivity disorder (ADHD; Ezell et al., 2019; R. J. Hagerman et al., 2018). Speech and language disorders are also common in this population, and decreased speech intelligibility is a prominent feature of the FXS communication profile (Abbeduto & Hagerman, 1997; Adlof et al., 2015; Barnes et al., 2009; Finestack & Abbeduto, 2010; Shaffer et al., 2020). Anecdotal reports describe perceived rapid rate of speech and speech that sounds “cluttered” in individuals with FXS (e.g., Hanson et al., 1986). However, cluttering has not been systematically examined in FXS, leaving SLPs with limited guidance on the assessment and intervention for cluttering in this group.

Although increased language disfluencies (i.e., repetitions, interjections, revisions and pauses) have been reported in individuals with FXS (Kover & Abbeduto, 2010; Van Borsel et al., 2008), the presence of cluttered speech in FXS has not been formally studied with the exception of one study including 10 males with FXS aged 3-8 years (Hanson et al., 1986). Investigators used a classification system that evaluated fluctuations in speech rate, rate of repetitions, history of articulation problems, attention problems, developmental delay, history of speech problems in a close relative, and self-awareness of a speech problem. These investigators found that 9 out of 10 participants exhibited a perceived rapid and irregular rate of speech with repetitions of sounds, words, or phrases. The most salient cluttering features present were perceived abnormal rate, repetitions, and atypical pauses.

Relationships Between FXS Phenotype and Cluttering

Underlying causal mechanisms for cluttering are unknown; however, emerging evidence suggests cluttering is related to a number of processes, including cognitive and language ability, and executive function (Myers, 2011; St. Louis & Schulte, 2011; Van Zaalen-Op’t Hof et al., 2009). Research in other neurodevelopmental disabilities such as Down syndrome has found that individuals with lower nonverbal IQ clutter or show symptoms of cluttering more often than individuals with typical development (Coppens-Hofman et al., 2013; Eggers & Van Eerdenbrugh, 2018; Van Borsel & Vandermeulen, 2009). Thus, general cognitive delay, as reflected by IQ may interfere with fluent speech and disrupt intelligibility. Increases in “normal” disfluencies and cluttering also occur in disabilities in which expressive language deficits, such as specific language impairment, are a feature (St. Louis & Schulte, 2011; Thordardottir & Ellis Weismer, 2002). Finally, an increase in the features of cluttering has also been associated with deficits in pragmatic language, executive functioning, and expressive language (Bangert & Finestack, 2020; Belser & Sudhalter, 2001; Jones et al., 2017; Navarro-Ruiz & Rallo-Fabra, 2001; Turkstra et al., 2004).

Cognitive and language delays are prominent features of the core FXS phenotype, and thus could contribute to cluttering in this population. Further, individuals with FXS show high rates of comorbid ASD, ADHD, and anxiety, which may increase vulnerability for cluttering. Individuals with nonsyndromic ASD and individuals with ADHD, for example, show high rates of cluttering and/or excessive disfluencies (Scaler Scott & St. Louis, 2009; Scaler Scott et al., 2014; Engelhardt, Ferreira, & Nigg, 2011; Redmond, 2004). Positive associations between anxiety and disfluent speech have been documented in the general population (Goberman et al., 2011; Iverach et al., 2014, 2016), although there are no studies that have directly investigated the association between anxiety and cluttering. In individuals with FXS, some research indicates that the incidence of increased rate and disfluent speech is triggered by anxiety associated with social-conversational demands (Belser & Sudhalter, 2001), although cluttering per se has yet to be examined in association with anxiety in this population. In a small-scale study, Kover and Abbeduto (2010) found reduced speech intelligibility in individuals with FXS who also had a co-occurring diagnosis of ASD. Improved understanding of how core features of the FXS phenotype relate to the presentation of cluttering in this group is needed to hone treatment approaches. For example, if executive dysfunction is associated with the presence of cluttering, this could spur future research examining whether integrating executive targets in treatments for cluttering in FXS result in improved outcomes (Myers, 2011).

Neurological Perspective of Cluttering in FXS

The study of cluttering in individuals with FXS also has the potential to provide insights about the mechanistic pathways that contribute to cluttering. There are currently no known genes implicated in the presence of cluttering, although there are well-documented neural signatures such as abnormal connectivity to and from the anterior cingulate cortex and supplemental motor area, cerebellum, and basal ganglia in cluttering and stuttering (Alm, 2011; Craig-McQuaide et al., 2014; Scott, 2006; Ward et al., 2015). Dysregulation in these regions, especially in the anterior cingulate cortex and supplemental motor area, may account for symptoms of cluttering. Current brain imaging data support the notion that the anterior cingulate cortex and supplemental motor area assemble speech by retrieving linguistic components from Wernicke's and Broca's areas, and are associated with volitional motor control, attention, suppression of automatic responses, and execution and timing of sequential behavior (Alm, 2011). The basal ganglia circuits facilitate word selection and filter out competing alternatives, and the cerebellum is involved in sequencing articulatory movements (Alm, 2011). Abnormal connectivity between these regions could impact speech and language processing, resulting in breakdowns in the flow of speech production. For example, disinhibition of the basal ganglia due to a hyperactive dopamine system may cause dysregulation of the medial frontal cortex, which includes the anterior cingulate cortex and supplemental motor area regions.

In FXS, many structures of the brain may be impacted, presumably due to *FMR1* plasticity-related protein synthesis deficiencies (Abbeduto & Hagerman, 1997; Hagerman et al., 2017). Alterations in the anterior cingulate cortex have been documented in individuals FXS

(Mercaldo et al., 2009), and these alterations could contribute to cluttering. Anatomical changes related to *FMR1* gene dysfunction have also been found in the posterior cerebellar vermis, which is a region important for processing sensory stimuli, sensory motor coordination, and motor output (Hagerman, 1997). Studies of adults with cerebellar disease or lesions in the posterior cerebellar vermis show increased dysarthria and reduced verbal fluency (Öztürk et al., 2014; Schmahmann & Sherman, 1998), and Ward et al. (2015) detected equal extensive activations of the vermis in adults who clutter and controls during a picture description task. Studying cluttering within the context of FXS, a monogenic condition that has relatively well understood genetic and neural mechanisms, could broaden our understanding of neuropathways involved in cluttering.

The Present Study

This is the first systematic study to characterize cluttering in males with FXS using objective measures of the LCD criteria and expert clinical opinion and to examine associations between cluttering and core aspects of the FXS phenotype. We focused on a well-characterized sample of young adult males with FXS, aged 18-26 years old. This study served to inform the prevalence, presentation, and associated features of cluttering in males with FXS. Our specific research questions and hypotheses were as follows:

1. What is the rate of cluttering in young adult males with FXS per expert clinical consensus following the LCD definition of cluttering? *We anticipated that 50-90% of males with FXS would demonstrate cluttering, based on previous findings of cluttering in individuals with FXS and other forms of intellectual disability* (Coppens-Hofman et al., 2013; Hanson et al., 1986).

2. What is the relationship between objective measurements of the cluttering features (speech rate, non-stuttering like disfluencies, atypical pauses, and over-coarticulation) and expert clinical consensus cluttering diagnoses? *We predicted that males with FXS who were identified as exhibiting cluttering per expert clinical opinion would show faster rates and increased frequency of the features of cluttering.*

3. Are core characteristics of the FXS phenotype (i.e., cognitive impairment, language impairment, anxiety, ADHD, and ASD symptoms) associated with cluttering diagnoses and/or objectively measured LCD features? *We predicted that phenotypic traits of FXS would be associated with increased likelihood of an expert clinical opinion cluttering diagnosis and with faster rates and increased frequency of the cluttering features.*

Method

Participants

Participants were 36 males with FXS, aged 18-26 years (*M* = 21.69 years, *SD* = 2.35) drawn from a larger, longitudinal study focused on language development in adolescent males with FXS (Abbeduto et al., 2019). FXS was confirmed via genetic testing (>200 CGG copies on 5'UTR of *FMR1*). Inclusionary criteria required that participants were native speakers of English and regularly used phrase speech (minimum 2- to 3-word utterances) according to caregiver report. Participants were also required to live at home with their biological mothers at study entry because the larger study included a focus on maternal factors. Participant demographic characteristics are presented in Table 1. All participants had an intellectual disability, with a mean nonverbal IQ of 39.54 (*SD* = 4.60) as found in the larger project (described below). Sixty-nine percent of the sample met the criteria for a comorbid ASD diagnosis (*n* = 24) on the Autism

Diagnostic Observation Schedule-2 (ADOS-2: Lord et al., 2012) and the Autism Diagnostic Interview-Revised (ADI-R; Rutter et al., 2003) using criteria laid out by Risi et al. (2006).

Participant recruitment was conducted nationally in the United States. Methods of recruitment included social media, word-of-mouth, family support groups, advertisements through the National Fragile X Foundation, and assistance from the Intellectual and Developmental Disabilities Research Center (IDDRC) Research participant registries of the Carolina Institute for Developmental Disabilities and the MIND Institute of the University of California Davis Health.

Table 1.

Participant Demographics

Variable		
Family Income		
	$20,000-$30,000	16.67%
	$30,001-$50,000	5.56%
	$50,001-$80,000	19.44%
	$81,001-$100,000	13.89%
	$100,001-$150,000	25.00%
	$150,001 or greater	19.44%
Race		
	Black or African American	5.71%
	American Indian or Alaskan Native	2.86%
	White	91.43%
Education		
	High school graduate	11.11%
	Some college	19.44%
	Associate's or technical degree	11.11%
	Bachelor's degree	38.89%
	Graduate degree	19.44%

Procedures

Assessments were conducted as part of a two-day research protocol that took place in a university research laboratory setting at the University of California Davis Health and the

University of South Carolina. The language and cognitive assessments were administered in a standardized order across both sites. A language sample was administered in the afternoon of the first day of the protocol. Caregivers were mailed a packet of questionnaires to complete in the two weeks prior to their study visit, which included the Childhood Behavioral Checklist **(**CBCL; Achenbach & Rescorla, 2000). Data for the present study were collected during the fourth yearly assessment of the larger longitudinal study, with the exception of the ASD assessment via the ADOS-2 and ADI-R which was completed only in the first yearly assessment in the larger longitudinal study. ASD symptoms in adolescents with FXS have been shown to be relatively stable over time (Hernandez et al., 2009). Procedures were approved by the Institutional Review Boards (IRB) of the respective university sites. Caregiver consent and participant assent were obtained.

Evaluation of Cluttering

Language Sampling Context

Cluttering was evaluated from language sampled from a 10-minute conversation with an examiner, following the standard procedures originally outlined by Abbeduto et. al (1995) and described in Kover et al. (2012). Conversation was elicited by a female examiner starting with an idiosyncratic topic of interest followed by a series of open-ended questions on standard topics (e.g., favorite sport, activities, family members). Examiners followed up with open-ended prompts to encourage participants to expand their talk (e.g., “Tell me why you like your friend so much”). The amount of time spent on each topic was driven by participant interest. The goal was to reach ten minutes of conversation with each participant; however, the conversation was considered over when the examiner exhausted all topics and prompts from the script. The average sample duration for the current study was 7.8 minutes (*SD*=2.03). The conversations took place in a quiet room and were recorded using digital Sony PCM voice recorders. To ensure consistency across occasions and examiners, all examiners completed rigorous fidelity training using the scripted content, including how, when, and how often they made comments and asked follow-up questions. The reliability protocol required practice administrations with children with typical development and children with developmental delay until 90% accuracy on a fidelity rubric of critical administration components was achieved (see Kover et al., 2012, for details). The complete manual describing the language sampling administration, training, and fidelity procedures can be access at *https://ctscassist.ucdmc.ucdavis.edu/ctscassist/surveys/?s=W9W99JLMNX* .

Cluttering Diagnosis via Expert Clinical Consensus

To determine the presence of cluttering via expert opinion, audio recordings of the language sample were independently reviewed for the presence of cluttering by two Ph.D.-level speech-language pathologists with expertise in the clinical evaluation of cluttering (K.S.S. and C.A.), each with more than 20 years of experience and one being board certified in fluency disorders. The samples were analyzed for cluttering according to the LCD definition of cluttering (Ward & Scaler Scott, 2011) and following the methods of Scaler Scott (2020). Samples identified as displaying cluttering needed to exhibit perceived rapid or irregular rate in connected speech at least some of the time. Per LCD criteria, in addition to a perceived rapid rate, a perceived excessive number of normal disfluencies, over-coarticulations, and/or atypical pauses were also required. The term “excessive” was defined as resulting in a perception of decreased efficiency of message transmission. Moments of over-coarticulation were distinguished from coarticulation due to other speech differences or disorders (e.g., articulation disorders, accent) by listening for patterns in speech across contexts (e.g., at different times in the sample). If there was a distorted, substituted, omitted sound, clinicians ascertained whether this pattern was observed repeatedly. For example, if distorted /r/ occurs elsewhere in the sample, over-coarticulation was ruled out and decreased intelligibility due to listener unfamiliarity with dialect or articulation difficulty was ruled in. Additionally, moments of over-coarticulation were confirmed when these moments were accompanied by rapid-sounding rate and by determining whether multisyllabic words in general were clearer when rate was slower for that particular sample. For participants for whom there was not initial agreement across the two experts, consensus was achieved via review of the samples and discussion.

Objective Characterization of Cluttering Features

Objective calculation of the presence of each of the LCD features was determined via coding of the audio-recorded language sample. These features were selected for inclusion because they align with the LCD definition of cluttering: non-stuttering like disfluencies (number of phrase repetitions, multisyllabic word repetitions, single syllable word repetitions without tension, revisions, interjections; Ambrose & Yairi, 1999; Yairi & Ambrose, 1992), the occurrence of over-coarticulation (i.e., deletion of one or more sounds or syllables of a word; Scaler Scott, 2020), and atypical pauses (pauses in places other than at syntactic boundaries; Scaler Scott, 2020). To correct for differences in sample length, the cluttering variables were converted to a percentage by dividing the total occurance of the variable by the total number of words, multiplied by 100. A rate variable reflecting average syllables per second was also calculated for each participant, using Prosogram (Mertens, 2004), a script for the speech analysis program Praat (Boersma & Weenink, 2016). Prosogram identifies the local intensity peaks in the filtered speech signal and then, using fundamental frequency, intensity, and variations in voicing in the spectrum, modifies the boundaries of the intensity peaks into syllable units (Mertens, 2004). Those units are then counted and divided by the total number of seconds for the sample.

Coding for the cluttering features was conducted by research assistants who had received at least 10 hours of training on the coding of fluency variables by a board-certified fluency specialist (K.S.S.). During training, coders transcribed and coded fluency features from five different videos of children and adults with cluttering and/or stuttering symptoms. In areas in which there was less than 90% agreement among raters, discrepancies were discussed among the raters and a cluttering specialist (K.S.S.), and criteria were refined as needed to increase objectivity of definition. Areas with less than 90% agreement were re-coded one week later, using (if applicable) revised objective criteria. Coders were determined to have completed the training after inter-rater reliability was ≥90% over three consecutive samples. Once coders completed reliability training, each sample was coded by one research assistant and then a second independent coder followed the original coder and double-scored the first 20% of utterances. If agreement between the two independent coders was <80% on observed cluttering features, a third independent coder evaluated the sample and all three coders achieved consensus via discussion, with the consensus scores used in analysis. Prior to consensus, inter-rater reliability was calculated with intraclass correlation coefficients (ICCs) following Koo and Li (2016), with values less than 0.50 considered poor reliability, 0.50-0.75 moderate reliability, 0.75-0.90 good reliability, and greater than 0.90 excellent reliability. Reliability was as follows: ICC (3, 5) = 0.93 for word repetitions, 0.82 for phrase repetitions, 0.88 for interjections, 0.80 for atypical pauses, 0.68 for over-coarticulations, and 0.60 for revisions. Thus, inter-rater reliability was “moderate” to “excellent” prior to consensus coding, and all samples that had poor reliability across raters were consensus coded.

Measures of Core Aspects of the FXS Phenotype

Nonverbal IQ

The Leiter-R (Roid, G., & Miller, 1997) is a nonverbal measure of intellectual functioning normed for individuals between the ages of 2-21 years. The Brief IQ was obtained, which consists of four subtests: figure ground, form completion, sequential order, and repeated patterns. For participants who were over the age of 21 years (*n* =17) we used the upper age limit of the Leiter-R norms (i.e., 21 years) to compute standard scores, consistent with Abbeduto et al. (2019) and Roberts et al. (2018). Given that all participants were in the moderate-severe impairment IQ range and performed at the floor or near the floor in terms of Leiter-R standard scores, growth scale values, which provide an index of absolute ability, were used instead.

ASD Symptom Severity

The ADOS-2 (Lord et al., 2012) calibrated severity score was used to detect the presence of ASD and determine the severity of ASD symptoms. In the ADOS-2, the examiner codes behavioral items related to communication, social interaction, imagination/play, stereotyped behaviors/restricted interests, and abnormal behaviors based on observations made during a semi-structured interaction with the participant. Calibrated severity scores range from 1 to 10, with a “1” indicating minimal-to-low ASD symptom severity and a “10” indicating high ASD symptom severity. All project staff examiners who administered the ADOS-2 were trained to research reliability standards. To establish cross-site reliability, 10% of videorecorded administrations were randomly selected for review and consensus scoring across sites. Consensus codes were reached for each scored item via group discussions and each examiner then calculated mean percent agreement with the consensus codes. Inter-rater agreement across items averaged 80%.

Anxiety and ADHD Symptoms

The Childhood Behavior Checklist **(**CBCL; Achenbach & Rescorla, 2000) Anxiety Problems and Attention Problems subscale raw scores were used to index symptoms of anxiety and ADHD. The CBCL is a standardized informant report measure used to identify emotional and behavioral problems in children between the ages of 6-18 years based on caregiver report. For the measure, primary caregivers rated their children on a 3-point scale, with 0=Not true, 1=Somewhat/sometimes true, and 2 =Very/often true on 113 items. DSM-oriented subscales for anxiety problems and ADHD symptoms were computed. The Anxiety Problems subscale is a six-item scale that represents symptoms of general anxiety, social anxiety, and specific phobia. The Attention Problems subscale is a 10-item scale that represents symptoms of ADHD including inattention, impulsivity, and hyperactivity. Higher scores indicate higher levels of difficulty in each area. Raw scores were used in analysis because not all participants fell within the age range for test norms. The 6-18 year-old form was used because the adult form was judged to be inappropriate for the developmental level of the sample, consistent with the methods of studies focused on young adults with FXS (Chromik et al., 2019; J. Roberts et al., 2019; J. E. Roberts et al., 2018). Caregivers completed the CBCL.

Language Ability

Mean length of utterance in morphemes (MLU) was calculated as an overall language ability measure. MLU is a widely used benchmark of early expressive syntactic language ability, and commonly used in individuals with language impairments (Condouris et al., 2003). MLU was calculated according to Systematic Analysis of Language Transcripts (SALT; Miller & Chapman, 2000) conventions, using transcripts that were transcribed from the audio files by trained research assistants. Transcription training consisted of analysis of practice language samples until >90% morpheme agreement among transcribers was achieved over three consecutive training files. Twenty-five percent of the participant's language samples were randomly selected and transcribed by a second independent transcriber to obtain ICCs (Koo & Li, 2016) for inter-rater reliability for MLU. A high degree of reliability was found across raters, with ICC (3, 5) = 0.94.

Data Analysis

Analyses were conducted using SAS 9.4 (SAS Institute Inc., Cary, NC, USA). First, descriptive statistics and variable distribution were examined. No corrections were determined necessary following visual inspection of study variables. The first research question regarding the rate of cluttering in males with FXS was addressed via descriptive statistics comparing those who were determined to exhibit cluttering per expert clinical opinion to those who did not exhibit cluttering. To better describe the profile of cluttering in FXS, the cluttering subgroups were compared on age, Leiter-R growth scale scores, ADOS-2 severity score, MLU, CBCL ADHD and anxiety scores via a series of general linear models with cluttering status as the primary predictor. Next, the second research question regarding the relationship between objective cluttering features and expert clinical opinion was addressed. A series of general linear models tested differences in the presentation of each of the seven objective cluttering features (percentages of phrase repetitions, word repetitions, revisions, interjections, over-coarticulations, and atypical pauses, and the number of syllables per second), across the cluttering subgroups. An FDR correction procedure was applied within each series of analyses to correct for multiple comparisons (Benjamini & Hochberg, 1995). The relationship between objective cluttering features and expert clinical cluttering diagnoses was also examined by analyzing the relative contributions of each of the objective cluttering features to the expert-opinion cluttering diagnosis. A single logistic regression model tested the objective cluttering features as relative predictors of the presence of cluttering via expert clinical opinion. To inform the specification of this logistic regression model and rule out collinearity among variables, a Pearson's correlation matrix was computed among the objective cluttering features (see Table 2). Finally, we addressed the third research question regarding the relationship between cluttering (objective features and expert clinical diagnoses) and core aspects of the FXS phenotype by computing Pearson's correlations between each of the objective cluttering features and the Leiter-R growth scale values, ADOS-2 severity score, CBCL ADHD subscale raw score, CBCL Anxiety subscale raw score, and MLU. A logistic regression tested these phenotypic features as predictors of the presence of cluttering as determined via expert opinion.

Table 2

Correlation Matrix

	1	2	3	4	5	6	7
1. Phrase Repetition Percent	1.00	.48*	.23	<.01	-.24	-.19	-.12
2. Whole Word Repetition Percent		1.00	.22	-.03	.34*	.17	-.28
3. Revision Percent			1.00	.40*	-.17	-.25	-.30
4. Interjection Percent				1.00	-.07	-.04	-.12
5. Over-coarticulation Percent					1.00	.50*	-.18
6. Atypical Pause Percent						1.00	-.40*
7. Syllables per Second							1.00

* $p<.050$

Results

Rates of Cluttering per Expert Clinical Consensus

The consensus of the clinical experts indicated that 50% of participants met LCD criteria for cluttering based on the characteristics exhibited in the language sample. There were no differences across subgroups of participants who did and did not meet criteria for cluttering in age (FDR-corrected $p = .780$, $R^2 = .01$), nonverbal cognitive ability as indicated by the Leiter-R growth scale value score (FDR-corrected $p = .713$, $R^2 = .03$), MLU (FDR-corrected $p = 0.825$, $R^2 < .01$), ASD symptom severity score (FDR-corrected $p = 0.825$, $R^2 < .01$), or scores on the CBCL Anxiety Problems (FDR-corrected $p = .714$, $R^2 = .04$) or ADHD (FDR-corrected $p = 0.416$, $R^2 = .09$) subscales. See Table 3 for descriptive statistics of the core developmental/behavioral measures of the FXS phenotype across the subgroups.

Table 3

Measures of Core Aspects of the FXS Phenotype across Cluttering Subgroups

	Group		
Variable	Subgroup without Cluttering (*n*=18)	Subgroup with Cluttering (*n*=18)	Full Sample (*n*=36)
Nonverbal IQ[a]			
M (*SD*)	39.18 (4.48)	39.89 (4.83)	39.54 (4.60)
Range	36.00-52.00	36.00-52.00	36.00-52.00
Leiter-R GSV[b]			
M (*SD*)	442.38 (9.09)	469.00 (8.97)	468.11 (9.01)
Range	441.00-485.00	453.00-484.00	441.00-485.00
Receptive Vocabulary[c]			
M (*SD*)	51.11 (15.78)	59.88 (19.57)	55.50 (18.08)
Range	20.00-86.00	20.00-94.00	20.00-90.00
ASD symptoms[d]			
M (*SD*)	5.22 (2.18)	5.53 (2.32)	5.37 (2.23)
Range	1.00-9.00	2.00-10.00	1.00-10.00
MLU			
M (*SD*)	7.22 (3.26)	7.00 (2.75)	7.11 (2.97)
Range	2.54-13.72	3.87-12.63	2.54-13.72
Total number of words			
M(SD)	874.83(394.44)	878.89(447.82)	876.86 (419.91)
Range	255.00-1481.00	302.00-1803.00	255.00-1803.00

Anxiety[e]			
M (*SD*)	3.44 (2.85)	2.39 (2.40)	2.92(2.66)
Range	0.00-8.00	0.00-9.00	0.00-9.00
ADHD[f]			
M (*SD*)	5.17 (3.22)	7.72 (4.80)	6.44(4.23)
Range	0.00-11.00	0.00-16.00	0.00-16.00

[a]Leiter-R Brief IQ scaled score, [b]Leiter-R growth scale values, [c]PPVT-3 standard scores [d]ADOS-2 Calibrated Severity Score; [e]Childhood Behavior Checklist (CBCL) anxiety subscale raw score; [f]CBCL ADHD subscale raw score.

Comparison of Objective Features across Cluttering Subgroups

A significant group effect was detected for phrase repetitions (F [1, 35] = 12.93, FDR-corrected $p = .007$, $R^2 = .28$), with phrase repetitions occurring more frequently in those who met criteria for cluttering via expert clinical opinion. The effect of group did not account for significant variance in whole word repetitions (FDR-corrected $p = .080$, $R^2 < .01$), revisions (FDR-corrected $p = .705$, $R^2 = .04$), interjections (FDR-corrected $p = .997$, $R^2 = .13$), over-coarticulations (FDR-corrected $p = .997$, $R^2 < .01$), atypical pauses (FDR-corrected $p = .952$, $R^2 = .02$), or syllables per second (FDR-corrected $p =.997$, $R^2 < .01$). See Table 4 for descriptive statistics of the objective cluttering features across the subgroups.

Table 4

Descriptive Statistics of Objective Cluttering Features across Cluttering Subgroups

	Group		
Variable	Subgroup without Cluttering (*n*=18)	Subgroup with Cluttering (*n*=18)	Full Sample (*n*=36)
Phrase Repetition Percent			
M (SD)	0.35 (0.32)	0.90 (0.57)	0.62 (0.53)
Range	0.00-1.09	0.37-2.36	0.00-2.36
Word Repetition Percent			
M (SD)	0.53 (0.51)	1.08 (0.88)	0.80 (0.76)
Range	0.00-1.88	0.00-3.18	0.00-3.18
Revision Percent			

M (SD)	1.01 (0.98)	1.45 (1.19)	1.23 (1.10)
Range	0.00-3.46	0.33-4.99	0.00-4.99
Interjection Percent			
M (SD)	3.56 (2.83)	3.25 (2.67)	3.41 (2.72)
Range	0.38-10.91	0.00-8.84	0.00-10.91
Over-Coarticulation Percent			
M (SD)	2.60 (5.14)	2.76 (5.98)	2.68 (5.49)
Range	0.00-4.02	0.00-20.91	0.00-20.91
Atypical Pause Percent			
M (SD)	0.64 (0.98)	0.43 (0.77)	0.53 (0.88)
Range	0.00-4.02	0.00-2.65	0.00-4.02
Syllables per Second			
M (SD)	4.34 (0.62)	4.34 (0.91)	4.34 (0.76)
Range	2.96-5.33	2.48-5.79	2.48-5.79

Relative Predictive Value of Objective Features in Expert Clinical Opinion Diagnosis

Increased frequency of phrase repetitions emerged as the only significant predictor of the presence of cluttering (χ^2 [1, n = 36] = 6.650, p = .010), with none of the other objective features accounting for significant variance in cluttering diagnosis via expert clinical opinion (all p's > .260); see Table 5 for odds ratios. For every unit increase in phrase repetition, the odds of meeting the LCD-definition criteria for cluttering increased by a factor of 165.19 (95% CI: 3.41, >999.99), see Figure 1.

Table 5.

Logistic Regression Model Showing Objective Cluttering Features as Relative Predictors of Cluttering Diagnosis via Expert Clinical Opinion

	Odds Ratio (CI 95%)	*p*-value
Phrase Repetitions	165.186 (3.408, >999.999)	.010*
Whole Word Repetitions	1.404 (0.190, 10.130)	.737
Revisions	1.419 (0.471, 4.274)	.534

Interjections	0.902 (0.643, 1.266)	.551
Over-Coarticulations	1.127 (0.915,1.387)	.261
Atypical Pauses	0.892 (0.224, 3.560)	.872
Syllables per Second	2.075 (0.496,8.679)	.318

*p <.050

Relationship between the FXS Phenotype and Cluttering Features and Diagnosis

Results of the logistic regression model indicated that symptoms of ADHD and anxiety symptoms were significant predictors of the presence of cluttering (χ^2 [1, n = 35] = .526, p = .011; χ^2 [1, n = 35] = -.842, p = .020, respectively), as was cognitive ability (χ^2 [1, n = 35] = .150, p = .044). Neither ASD severity (χ^2 [1, n = 35] = .040, p = .862) nor language ability as indicated by MLU (χ^2 [1, n = 35] = .068, p = .715) were significant predictors of the presence of cluttering. For every unit increase in ADHD symptoms, the odds of meeting the LCD-definition criteria for cluttering increased by a factor of 1.69 (95% CI: 1.13, 2.53), see Figure 2. For every unit increase in anxiety symptoms, the odds of meeting the LCD-definition criteria for cluttering decreased by a factor of 0.43 (95% CI: 0.21, 0.87), see Figure 3. For every unit increase in cognitive ability, the odds of meeting LCD-definition criteria for cluttering increased by a factor of 1.16 (96% CI: 1.00-1.34), see Figure 4. Correlations between the objective cluttering features and the FXS phenotypic variables were also examined and are presented in Table 6.

Discussion

The current study investigated cluttering in a well-characterized sample of young adults with FXS and examined relationships between cluttering and aspects of the FXS phenotype. Findings showed that 50% of individuals presented with cluttering per clinical expert opinion.

Cluttering was more likely to be present when individuals with FXS exhibited an increased use of phrase repetitions but was not predicted by other linguistic features that were objectively measured from language samples, including whole word repetitions, revisions, interjections, over-coarticulations, atypical pauses, and fast rate. Although the presence of cluttering was not associated with ASD symptoms or language ability, cluttering was more likely to occur when ADHD symptoms were elevated. Surprisingly, increased anxiety and decreased nonverbal cognitive skills were associated with reduced likelihood of cluttering. Overall, this study contributes to our understanding of the clinical presentation of cluttering in young adult males with FXS and may lead to improved understanding of the potential underlying mechanisms of cluttering and eventual refinements to the treatment and diagnosis of cluttering in FXS.

Prevalence of Cluttering in Young Adults with FXS

Half of the young adult males with FXS in the current study were identified as having a cluttering disorder by clinical experts. This percentage is consistent with that observed in individuals with other forms of intellectual disability (Coppens-Hofman et al. 2013). Yet, it is notably lower than the rate reported in a preliminary study by Hanson et al. (1986), in which nine of a sample of ten children with FXS (aged 3-10 years) presented with cluttering. The increased rate in the Hanson report may be due to the younger age of the sample, as younger children may experience developmental spurts in speech and language learning which may impact fluency (Scaler Scott & St. Louis, 2009). Additionally, we applied the more narrow LCD criteria to identify the presence of cluttering, which is the currently the most widely accepted characterization of cluttering (St. Louis & Schulte, 2011; Ward & Scaler Scott, 2011); these diagnostic criteria had not yet been developed at the time of the Hanson study and likely yielded a more accurate characterization of cluttering in the present report. Of course, the very small sample of Hanson et al. also decreases confidence in the prevalence observed. The high rates of cluttering detected in this report, with 50% of males with FXS affected, suggest the need for increased awareness of risk for cluttering in FXS among SLPs which may result in improved selection of intervention targets to increased communication efficiency in individuals with FXS. This study also highlights the important need for increased research in intervention efforts targeting cluttering in this population.

The high percentage of cluttering in our study also suggests the need for future research on potential common genetic and neural pathways implicated in cluttering and FXS. Given our results indicating that FXS is highly enriched for cluttering, it is possible that *FMR1* plays a role in the manifestation of cluttering. There are currently no known genes associated with cluttering, although studies suggest that cluttering shows substantial heritability at a rate of .53-.65 (Fagnani et al., 2011). *FMR1* and its product, FMRP, have widespread involvement in neurocognitive development (Hagerman & Hagerman, 2002; Hagerman, 1997; Loesch & Hagerman, 2012; Tassone et al., 2000) and could potentially play a mechanistic role in cluttering. Indeed, variability in the CGG repeat length on *FMR1*, including CGG repeat lengths falling within the normal range, have been connected with more disfluent speech (Klusek et al., 2018). More research into the role of *FMR1* in cluttering could be beneficial in informing mechanistic pathways underlying the condition.

Relationship between Objectively Measured Cluttering Features and Expert Clinical Opinion Diagnoses

In addition to characterizing rates of cluttering per the LCD criteria applied by expert clinical opinion, this study also utilized objectively measured cluttering features from the language samples in order to systematically characterize cluttering. Of the cluttering features measured (e.g., phrase and word repetitions, revisions, interjections, over-coarticulations, atypical pauses, and syllables per second), phrase repetition was the only feature that was predictive of the presence of cluttering, and the only feature that distinguished the subgroups of those with and without cluttering. Phrase repetitions, which have been primarily studied in FXS as an aspect of perseverative language, are a feature of the FXS language profile that has been well-documented in previous studies (Abbeduto et al., 2007; Hanson et al., 1986; Martin, 2009; Sudhalter & Belser, 2001; Van Borsel et al., 2008). This study builds on our understanding of the use of phrase repetitions in FXS by establishing their association with cluttering, although it remains unclear why this aspect of language was specifically linked with cluttering. One possibility is that phrase repetitions in FXS may be associated with unique paralinguistic features that lead to the perception of cluttered speech (for example, rapid rate exhibited during phrase repetitions); this hypothesis may be explored in future studies. Evaluating the presence of and targeting phrase repetitions could be a useful tool for SLPs diagnosing and treating cluttering, although more research is needed. For example, does treatment aimed toward slowing rate and reformulating utterances reduce the frequency of phrase repetitions and result in reduced severity of cluttering or improved intelligibility?

Aside from phrase repetitions, no other objectively measured cluttering features significantly predicted cluttering status, nor did those features differ across the subgroups of those who were classified as clutterers and those who were not. This was a surprising finding, especially for our measure of rate given that rapid or irregular rate is the only mandatory characteristic of cluttering in LCD criteria. Increased rate of speech has been noted clinically in FXS and was thought to relate to disfluencies and reduced intelligibility in this group (Belser & Sudhalter, 2001; Mirrett et al., 2003; National Fragile X Association, 2020). Therefore, we expected a faster rate to be particularly notable among males with FXS who exhibited cluttering. Although abnormal rate was noted subjectively by the clinical experts who evaluated the presence of cluttering, this finding was not observed in our objective measure of rate. Indeed, the mean rate overall was 4.34, which seems to fall within normal limits for average American English speaking adults (Jacewicz et al., 2009). Nonetheless, these findings do align with a number of previous studies of cluttering outside of the context of FXS that have found no rate differences across individuals who clutter and do not clutter (Bakker et al., 2011; Garnett & St. Louis, 2014; Lees et al., 1996; Van Zaalen- op 't Hof et al., 2009). It has been hypothesized that the rate in cluttering may be perceived as rapid due to variation in rate affected by other cluttering symptoms (Williams, 2019). This hypothesis may align with the high frequency of repetitions as a distinguishing feature of cluttering in the current sample. In the case of FXS, it is also possible that fast speech rate is a central aspect of the communication phenotype and is not unique to those who clutter. Future research should explore the inclusion of other methods for quantifying rate that may better align with the perception of fast speech.

Associations Between FXS Phenotype and Cluttering Diagnosis/Symptoms

Increased ADHD symptoms was a significant phenotypic predictor of the presence of a cluttering disorder in our sample of individuals with FXS. This finding aligns with evidence suggesting that deficits in executive functioning that are commonly observed in ADHD, specifically organization and planning, are associated with an increase in language disfluencies (Oomen & Postma, 2001; Turkstra et al., 2004). This also aligns with self-reports and reports of cluttering and symptoms of ADHD in adults with cluttering (St. Louis & Scaler Scott, 2011; David Ward & Scaler Scott, 2011). Future investigation of specific executive functioning abilities associated with cluttering in individuals with FXS could inform the relationship among these variables and potentially spur future intervention studies aimed toward improving cluttering via targeting executive functions. Of note, SLPs should be aware that individuals with FXS and co-occurring ADHD may be at higher risk for cluttering or conversely, individuals with FXS who clutter may benefit from screening for ADHD co-occurrence.

Contrary to our predictions, *lower* anxiety significantly predicted the presense of cluttering. Anxiety and disfluent speech are associated the general population (Goberman et al., 2011; Iverach et al., 2014, 2016). Specifically within FXS, anxiety and associated hyperarousal during social interactions have been theorized to underlie repetitive speech (Belser & Sudhalter 2001). However, our findings suggest that higher anxiety levels result in fewer instances of cluttering. This could reflect an increase in self-monitoring due to anxiety, which aligns with the notion that for many with cluttering, symptoms "normalize" when the speaker is recorded due to increased self-monitoring (Van Zaalen-op & Reichel, 2015). The measurement of anxiety in individuals with intellectual disability is imprecise, and it is also possible that associations were skewed by under-reporting of symptoms in more affected individuals (Cordeiro et al., 2011). More research in this area is needed. Future studies incorporating physiological measures of stress may inform the manifestation of cluttering. For example, measures of heart rate or heart rate variability during a social interaction could inform whether cluttering is associated with physiological hyperarousal, which is a hallmark feature of FXS (Klusek et al., 2015). Currently no empirical studies examining the link between arousal and disfluent speech or cluttering in FXS exist.

Another somewhat surprising finding was that higher nonverbal cognitive ability was associated with a higher likelihood of cluttering. The reason for this pattern is unclear. One potential explanation could be that increased cognitive ability translates to better language skills and increased speech verbosity, which in turn allows for more opportunities to exhibit cluttering. However, this seems unlikely considering that our analysis controlled for MLU. It is notable that the range of cognitive skills was restricted in our sample (IQ's ranged from 36-53) and therefore the detected pattern may not generalize across the full range of intellectual function. Follow-up studies involving females with FXS, who exhibit greater variability in cognitive skills than males, may be useful in untangling the relationship between cognitive impairment and cluttering in individuals with FXS. Nevertheless, the relationship between cognitive performance and cluttering has clinical implications regarding the identification of co-occurring conditions in individuals with intellectual disability. For example, clinicians may be less likely to recognize and treat cluttering when it occurs within the context of a genetic syndrome such as FXS. This phenomenon is known as *diagnostic overshadowing*, or when symptoms are attributed to intellectual disability rather than being recognized a separate co-morbidity, and is noted in the literature as problematic (Jopp & Keys, 2001; Mason & Scior, 2004; Reilly et al., 2015). Our findings suggest that the opposite it true -- cluttering was more likely to occur in males with FXS when cognitive skills were less impaired. Therefore, cluttering should be recognized as a co-occurring condition in FXS that may warrant intervention.

Finally, MLU was not associated with increased cluttering. Although MLU was employed in the current study as a measure of overall language ability, MLU may also be conceptualized as an indicator of linguistic complexity, particularly when MLU is low (Condouris et al., 2003; Scarborough, 1990). Increased linguistic complexity has been associated with greater disfluency in previous literature (Maclachlan & Chapman, 1988; Wagner et al., 2000) and we therefore might have expected higher MLU to relate to increased features of cluttering. However, only phrase repetitions were significantly associated with MLU, and the relationship was *negative,* where phrase repetitions increased at lower MLU's. Thus, we did not observe increased cluttering features at higher linguistic complexity, as marked by MLU. The inclusion of other complexity measures in future studies may be useful in delineating the relationship between linguistic complexity and cluttering.

Strengths and Limitations

This study is the first to examine cluttering in young adults with FXS. A particular strength is that cluttering was evaluated by clinical professionals with over 20 years of combined experience in diagnosing cluttering, applying the current LCD definition of cluttering. We also include well-defined objective measurements of cluttering characteristics from language transcripts. The sample of individuals with FXS was homogenous in age and cognitive and language ability allowing for a more controlled sample. This was done in part by only including males; however, we acknowledge that larger scale studies involving females with FXS are needed. We also note our sample was 90% white, and although there was a range of economic backgrounds represented in the sample over half of the participants came from households with an income of over $80,000. It is therefore unclear how generalizable findings are to more diverse groups. Lack of diversity is a frequent problem in FXS research (Riley et al., 2017), and future studies should increase efforts to recruit families from underrepresented populations. A final limitation is that cluttering was determined from a 10-minute speech sample in a highly controlled environment. The use of a longer sample or less controlled context may have allowed for the expression of increased or more variable features of cluttering. However our use of a 10-minute sample is similar to the sample lengths used in previous studies (e.g., Coppens-Hofman et al., 2013; Eggers & Van Eerdenbrugh, 2018; Scaler Scott et al., 2014; Sudhalter & Belser, 2001). We also note that cluttering may or may not be influenced by individual examiner's behavior, such as asking more questions or making statements. We did attempt to control for this by providing extensive training to examiners and following a standardized script, however analyses of the amount and type of talking done by the examiner and its relationship to cluttering is another possible future direction.

Conclusions

Clinicians who work with individuals with FXS should be aware that this genetic diagnosis confers heightened risk for cluttering. As many as half of individuals with FXS may be affected by cluttering, and aspects of the FXS phenotype such as ADHD and anxiety symptoms may play a role in its presentation. Given the significant consequences of cluttering on interpersonal relationships and ability to communicate effectively at school or work, this study highlights the need for more research on cluttering in FXS, including intervention work.

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Figures

Figure 1

Probability of Cluttering Diagnosis by Phrase Repetition Percent with 95% Confidence Limits

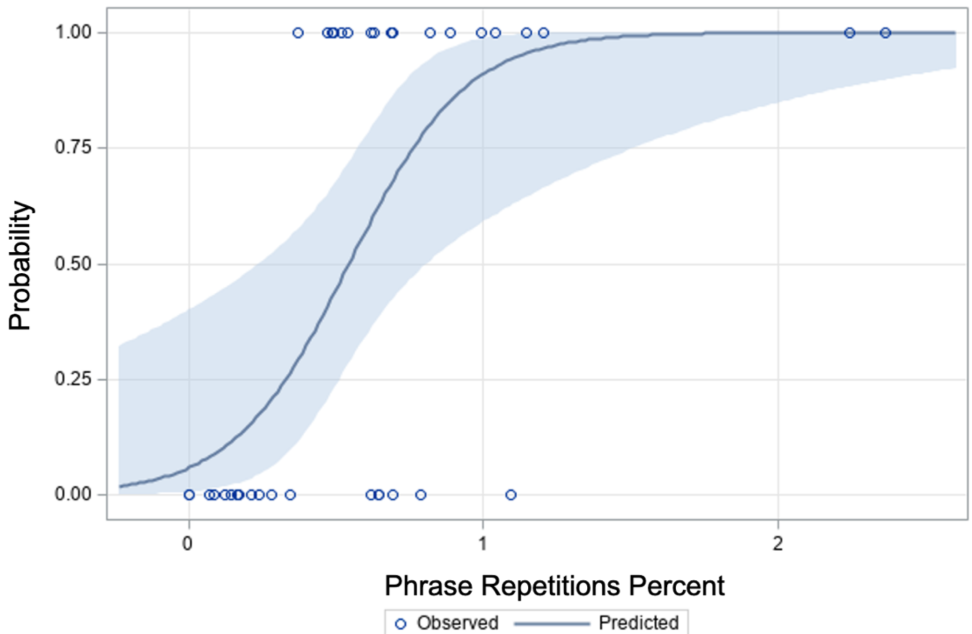

Figure 2

Probability of Cluttering Diagnosis by ADHD Symptoms with 95% Confidence Limits

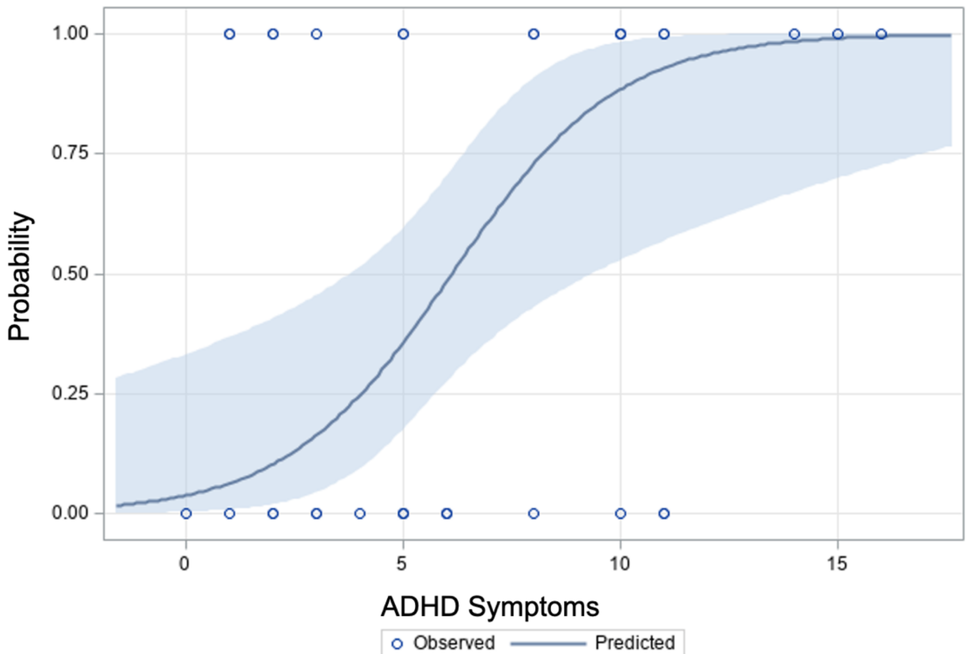

Figure 3

Probability of Cluttering by Anxiety Symptoms with 95% Confidence Limits

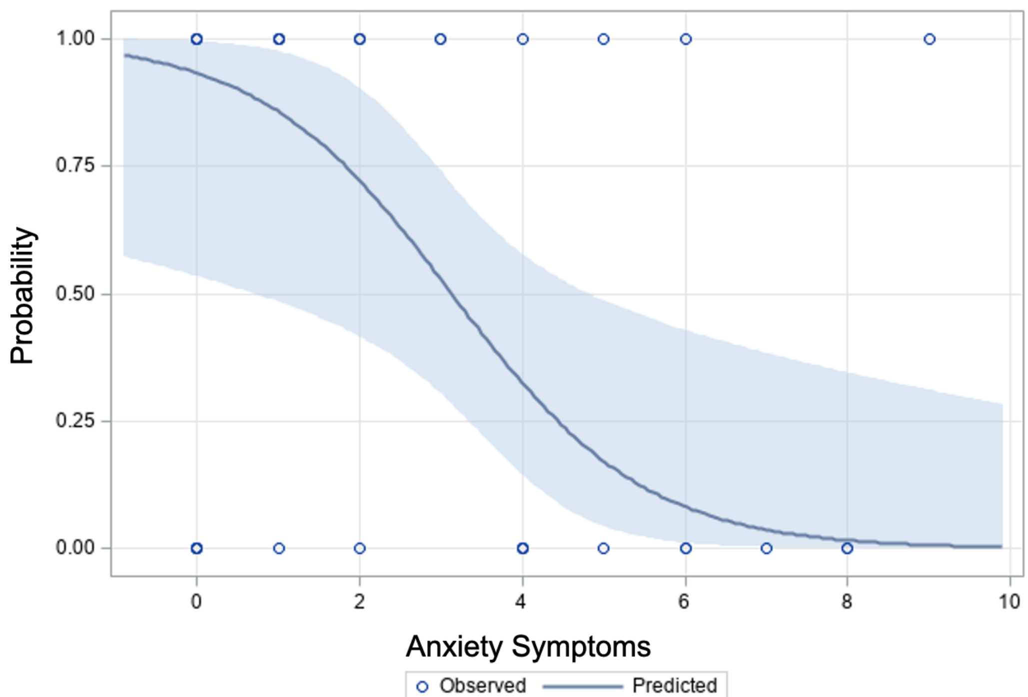

Figure 4

Probability of Cluttering by Cognitive Ability with 95% Confidence Limits