Maternal Requests and Child Compliance during Mother-Child Interactions in Adolescents with Fragile X Syndrome or Autism

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Abstract

Requests for action are used to ensure an action is completed. Parents use requests for action expecting that their child will comply with their requests. However, children are not always compliant. Noncompliance may be due to difficulty comprehending the request, especially when the request is complex or does not meet social expectations. Requests can vary in three different properties: directness, transparency, and conventionality. For instance, parents may use indirect requests (e.g., “Do you mind putting away the dishes?”), ambiguous requests (e.g., “Next we open the bag.”), or unconventional requests (e.g., “It would be great if the dishes were put away.”). Individuals who have cognitive, linguistic, and social delays or impairments, such as those with fragile X syndrome (FXS) or autism spectrum disorder (ASD), may have difficulty interpreting such requests. The general purpose of this study was to examine the extent to which language, nonverbal cognition, social skills, autism symptoms, and request properties predicted compliance with maternal requests for action by adolescent males with FXS or ASD.

Thirty-six adolescent males with FXS and their biological mothers participated in an ongoing longitudinal study, and an additional two participants with ASD and their mothers were recruited. Adolescents ranged from 12 to 18 years old. With their mothers, the adolescents completed a 10-minute snack task which was videotaped. Each video was coded behavior-by-behavior and from this, maternal requests for action and child compliance were assessed. Additionally, adolescents completed assessments of language and their mothers completed a parent-report interview of their son’s adaptive behavior. Autism symptomology and nonverbal cognition were also assessed in the FXS group.

The effects of request property, language, nonverbal cognition, social skills, and autism symptomology were considered using a series of multilevel regressions. Results indicated that

language ability and nonverbal cognition were strongly associated with compliance, as were social skills. Request properties were not associated with compliance. Autism symptomology was also associated with compliance such that individuals with higher autism symptom severity were less likely to comply. The two participants with ASD performed similarly to the FXS group on all measures.

Adolescent males with FXS or ASD were largely compliant with maternal requests for action regardless of request directness, transparency, or conventionality. Although the participants demonstrated high levels of compliance, individual differences in language, nonverbal cognition, and social skills accounted for some variation in compliance. Findings suggest that adolescent males with FXS who have lower language or social skills or who have elevated autism symptomology are less compliant. Additional adolescent characteristics and/or contextual demands may further predict compliance, so future research will need to identify and examine other potential predictors. This research highlights the ways in which parent language use may impact child behavior and demonstrates that there is no gold standard way for mothers to present requests that will promote compliance in adolescent males with FXS or ASD.

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Chapter I: Introduction

Requests for actions are an important parenting strategy through which parents can relay information about social expectations, promote behaviors of daily-living, and ultimately, foster independence in their children. For parents, requests can be used to encourage their children to engage in new or continuing positive or adaptive behaviors, or they can be used to terminate unwanted or maladaptive behaviors. However, when parental requests are unclear or overly-complicated, children may struggle to comply with them. Failure to comprehend parental requests for action could result in noncompliance and in turn lead to communication breakdowns, frustration, and maladaptive behaviors. Difficulty comprehending complex requests may be particularly evident for children with linguistic and cognitive delays and impaired social skills, such as boys with fragile X syndrome (FXS) or autism spectrum disorder (ASD), who have difficulty with social communication. Thus, it is preferable that parents of boys with FXS or ASD utilize parenting strategies that are appropriate for their child's age and social communication skills. This project examined the associations between maternal request style, maternal request complexity, child language and nonverbal cognition, child social skills, and child compliance with maternal requests for action in adolescent males with FXS or idiopathic ASD.

Fragile X Syndrome and Autism Spectrum Disorder

Fragile X syndrome and ASD are both neurodevelopmental disorders that can have profound impacts on an individual's communication, social skills, emotion regulation, and adaptive behavior. Fragile X syndrome is caused by excessive CGG trinucleotide repeats on the non-coding region of the *FMR1* gene, which is located on the X chromosome (Verkerk et al., 1991). *FMR1* produces the Fragile X Mental Retardation Protein (FMRP) which has been

implicated in neuronal differentiation, synaptogenesis, and neuronal plasticity during early neural development in utero (see further Colak et al., 2014). The elongated CGG repeat sequence alters production of FMRP, and partial or complete loss of FMRP production may alter synaptic plasticity thus affecting learning and memory (O'Donnell & Warren, 2002). Fragile X syndrome is an X-linked disorder, so males generally have more severe symptoms than females. Unlike FXS, idiopathic ASD does not currently have a known etiology, although it is believed that ASD may be heritable within some families (Sasson et al., 2013). Like FXS, ASD may also differentially affect the sexes, with three to four times more males than females receiving diagnoses (Loomes et al., 2017).

Although varied, males with FXS usually present with impairments in nonverbal cognition (Schneider et al., 2009), language (Abbeduto et al., 2007; Finestack et al., 2009), social skills (Roberts et al., 2002), and adaptive behavior (Klaiman et al., 2014). Males with FXS also demonstrate elevated autism symptomology (Abbeduto et al., 2019; Haebig et al., 2020) and elevated levels of maladaptive behaviors (Hall et al., 2016; Hardiman & McGill, 2018). An individual is diagnosed with ASD if they have persistent deficits in social-emotional reciprocity and nonverbal communication, difficulty with social relationships, and elevated rates of repetitive behaviors and restricted interests such as stereotyped behaviors, resistance to change, highly restricted, fixated interests, and/or unusual interest in sensory aspects of the environment (American Psychiatric Association, 2013). Unlike FXS, the profile of symptoms and deficits in ASD varies, as some, but not all, individuals with ASD have impairments in speech, language, and nonverbal cognition (Carpenter, 2013; Sigman & McGovern, 2005). Like FXS, many individuals with ASD present with elevated rates of maladaptive behaviors (American Psychiatric Association, 2013). Because males with FXS have known deficits in language and

social skills, in addition to elevated levels of problem behaviors, they constitute a group in which studying maternal requesting and child compliance may have important clinical implications. Similarly, due to their difficulty with social communication and elevated levels of problem behaviors, males with ASD also represent a unique population in which to study maternal requesting and child compliance.

Language and Cognitive Ability in Males with FXS or ASD

It has been robustly demonstrated that males with FXS have deficits in receptive language relative to age expectations. Roberts et al. (2001) demonstrated that young boys with FXS between 20 and 86 months of age gain receptive language skills at less than half the rate of normed expectations. Although they may eventually close the performance gap with mental age (MA) matched same-sex typically developing (TD) peers, males with FXS likely never reach chronological age (CA) expectations. Young boys with FXS perform similarly to MA expectations on measures of receptive vocabulary (Roberts et al., 2007), while young boys with FXS and comorbid ASD perform at or below MA expectations. As a group, young boys with FXS (regardless of autism symptomology) perform worse than MA matched TD boys on measures of receptive morphosyntax (Price et al., 2007). McDuffie et al. (2012) and Price et al. (2007) suggest that high autism symptomology may be related to increased impairment in receptive language in school-age and adolescent males with FXS.

In addition to impaired receptive language, the onset of speech, development of expressive vocabulary, and use of expressive morphosyntax are delayed in males with FXS (Brady et al., 2006; McDuffie et al., 2012; Philofsky et al., 2004; Price et al., 2008; Roberts et al., 2007; Sterling, 2009). Early in development, young boys with FXS (regardless of autism symptomology) are delayed in producing their first words (Brady et al., 2006; Hinton et al.,

2013) and demonstrate delays in early speech skills, specifically consonant inventory and syllable structure accuracy (Fielding-Gebhardt & Warren, 2019; Roberts et al., 2005). As first words and phonological processes develop into a growing lexicon, there remains a marked delay in the acquisition of expressive vocabulary in young boys with FXS (Roberts et al., 2001; Roberts et al., 2007). Males with FXS struggle with expressive morphosyntax, as do males with FXS and comorbid ASD (Price et al., 2008; Sterling, 2018). This delay, in combination with a reduced rate of acquisition (Roberts et al., 2001), suggests that males with FXS may never reach CA norms in expressive language skills.

Males with ASD may also demonstrate delayed acquisition of receptive and expressive language (Kover et al., 2013; Yoder et al., 2015), although this delay is not as prevalent as in FXS. In a sample of 49 boys with ASD (ages 4 to 11), Kover et al. (2013) reported deficits in receptive and expressive language ability and growth relative to TD peers. Boys and adolescent males with ASD may have higher language abilities than boys and adolescent males with FXS and comorbid ASD (Haebig & Sterling, 2017). Thus, although males with ASD often have difficulties with receptive and expressive language, their level of ability is heterogeneous and is likely to vary based on nonverbal cognitive ability (Kover et al., 2013; Sigman & McGovern, 2005).

Nonverbal cognition is also impaired across the lifespan in individuals with FXS. Males with FXS demonstrate impairments in visual-motor coordination, spatial memory, and arithmetic, and have overall low nonverbal IQ (Schneider et al., 2009). Lower cognitive performance has been noted in males with FXS and ASD compared to males with FXS only (Meguid et al., 2012). Numerous studies have demonstrated that nonverbal cognition accounts for significant variance in language outcomes in FXS (McDuffie et al., 2012; Roberts et al.,

2001; Roberts et al., 2007). Unlike FXS, not all individuals with ASD have impairments in nonverbal cognition given the highly heterogenous nature of the disorder. Rather, individuals with ASD may demonstrate unique profiles of strengths and weaknesses across cognitive domains (Kuschner et al., 2007). As in FXS, nonverbal cognition is associated with receptive and expressive language abilities in ASD (Kover et al., 2013).

Social Communication in Males with FXS or ASD

The ability to judge the appropriateness of language in a given context is an important social skill that may be impaired in individuals with FXS or ASD (Klusek et al., 2014; Losh et al., 2012). Klusek et al. (2014) reported that 10 to 12 year-old males with FXS perform worse on naturalistic and structured assessments of pragmatic language than MA expectations. Adolescent males with FXS use a high proportion of repetitive utterances during conversation (>25%) and may be dependent on rote phrases or sayings to carry on a conversation (Murphy & Abbeduto, 2007). Individuals with idiopathic ASD may fail to correctly use and identify emotion terms, gestures, contextual cues, and social initiations, and may struggle to maintain topics and participate in reciprocal conversation (APA, 2013). They may also rely on rote conversational devices and perseverate on topics or preoccupations (APA, 2013). Males with FXS or ASD may have difficulties with topic shifting/maintenance and may use atypical intonation patterns and scripting (Klusek et al., 2014).

School-age boys with FXS and comorbid ASD perform significantly worse than those with FXS and low autism symptomology on pragmatic language skills (Klusek et al., 2014; Losh et al., 2012). They also perform worse than TD control groups (Klusek et al., 2014; Losh et al., 2012), suggesting that an impairment in social use of language is part of the autism phenotype in males with FXS. This phenotype characteristic is reasonable to assume given that deficits in

social communication are a hallmark symptom of ASD. Indeed, when comparing pragmatic skills in school-age boys with ASD versus those with FXS and high autism symptomology, no difference is found (Klusek et al., 2014; Losh et al., 2012).

Social Language

The social use of language is an essential skill that is used in many contexts throughout one's daily activities. Knowledge of the social rules and subtleties associated with language use is critical for successful school, work, and personal relationships. Social use of language relies on adequate expressive and receptive language ability. As such, when these domains of language are impaired, it is reasonable to expect that pragmatic skill will also be impaired because social interaction relies on the ability to competently produce and comprehend language (Berko Gleason & Ratner, 2009). Within both FXS and ASD, additional investigations are necessary to determine exactly where deficits in pragmatic skills lie, their relationship with autism symptomology, and potential ways to target these deficits during therapeutic treatments.

One important pragmatic skill is the ability to infer meaning from a conversational partner's non-literal utterance. Inferring meaning from non-literal language may be particularly difficult for individuals with elevated autism symptomology, as individuals with ASD often struggle to interpret non-literal or figurative language (Carpenter, 2013; Kalandadze et al., 2016). Non-literal utterances are those in which there is a discrepancy between the semantics or linguistic meaning (the locutionary force) of the utterance and the intended meaning (illocutionary force) of the utterance (Kissine et al., 2012; Searle, 1969). The illocutionary force of an utterance determines the function of that utterance (e.g., comment, request, question, etc.), but may not align with the locutionary force or morphosyntax and semantics of the utterance (e.g., declarative, imperative, interrogative, etc.; Kissine et al., 2012). When this occurs, the

utterance is considered an indirect speech act. For example, an utterance such as (1) may be interpreted with its literal or direct meaning (i.e., *the room is nice and warm*), or with its non-literal or indirect meaning (i.e., *it is too warm in the room, so go open a window*).

(1) It's warm in here.

Listeners must be able to use contextual clues in combination with social information to determine whether the speaker intended the literal or non-literal meaning (Ervin-Tripp & Gordon, 1986; Kissine et al., 2015).

Requests for Action

The ability to correctly interpret non-literal utterances becomes particularly relevant when in the context of indirect requests for action. Requests for action are given so that a listener complies and completes the action (Ervin-Tripp & Gordon, 1986), but they vary in directness or the degree to which the intended meaning is explicitly stated in the utterance. Direct requests are imperative statements as in (2) and (3), where the intended meaning of the utterance (request) aligns with the morphosyntactic and semantic structure of the utterance (imperative). Indirect requests take many forms, such as (4) through (8) in which the intended meaning of the utterances (requests) do not match the morphosyntactic or semantic structures of the utterances (interrogatives and declaratives), see Table 1. Indirect requests may be confusing for the listener because they do not convey a literal meaning (Pinker et al., 2008). Rather, the request is veiled and the listener must comprehend and interpret the underlying meaning.

Table 1:

Example Requests for Action and Their Properties

	Locutionary Force	Illocutionary Force	Directness	Transparent	Conventional
(2) Stir the pudding.	Imperative	Request	Direct	Yes	Yes
(3) You need to stir the pudding.	Imperative	Request	Direct	Yes	Yes
(4) Can you stir the pudding?	Interrogative	Request	Indirect	Yes	Yes
(5) The pudding needs to be stirred.	Declarative	Request	Indirect	No	No
(6) I'd like you to stir the pudding	Declarative	Request	Indirect	Yes	No
(7) Would you mind stirring the pudding?	Interrogative	Request	Indirect	Yes	No
(8) Let's stir the pudding.	Declarative	Request	Indirect	No	Yes

Indirect speech acts have been widely considered in the literature (Clark, 1979; Ledbetter & Dent, 1988; Pinker et al., 2008; Rimac, 1985). Pinker et al. (2008) addressed the puzzling rationale for using indirect speech, stating that “indirect speech is inefficient, vulnerable to being misunderstood, and seemingly unnecessary” (p. 833). As the authors highlight, indirect speech is unnecessary because individuals are usually capable of interpreting the direct or literal meaning from a direct request. However, indirect requests are used, and often preferred, due to politeness norms in language and culture. Indirect requests may produce more comfortable interactions because they are perceived as more respectful. Additionally, they reflect the expected relationship between the speaker and listener, and they may help speakers portray social intelligence (Pinker et al., 2008).

Not only do requests for action vary in directness, they also vary in transparency and conventionality (Ervin-Tripp & Gordon, 1986). Transparency refers to the surface structure

specification of the agent, action, and (optional) object, while conventionality refers to the social-expectations and social-acceptability of the directive intent of the request (Ervin-Tripp & Gordon, 1986).

Transparency, akin to morphosyntactic structure, can be categorized as transparent or ambiguous. Every request necessarily contains a main verb accompanied by a subject. The main verb in the request requires, at minimum, an agent which is the entity that performs the verb. The assignment of an agentic relationship specifies the initiator of the action, also referred to as the subject (Carnie, 2013). So, in "Mary stirs the pudding", *Mary* is the agent, *stirs* is the main verb, and *the pudding* is the object. If the agent and main verb (and object) are overtly specified, or strongly implied, then the structure is transparent, as in (2) and (3) (note that English allows for an elided subject in imperative statements, such as "[you] Go to your room"). However, if the agent is ambiguous, unspecified, or not strongly implied, then the structure is no longer transparent and may be harder for the listener to interpret (Ledbetter & Dent, 1988). In (8) the subject is ambiguous as it can refer to the speaker, the listener, or both. Thus, (8) would be considered an ambiguous request. Critically, a transparent request will specify the action and who needs to complete the action (usually the listener) in such a way that the listener understands the requested behavior and the agent (Clark, 1979).

Conventional means of requesting adhere to the social norms of a culture which are utilized to ensure that the listener interprets the request with the correct illocutionary force (Clark, 1979). The conventionality of requests derives from the social information and context of the request. Ervin-Tripp and Gordon (1986) suggest that the social information in requesting includes the relationship between speaker and listener, the context of the request, and the attitude and mood of the speaker and listener. Indeed, this social information is required for

comprehension of the request (Ervin-Tripp & Gordon, 1986). The presupposed social relationships, in combination with the joint interpretation of contextual demands and the friendliness of the speaker and listener are realized upon correct interpretation of and compliance with the request. Thus, if the appropriate social information is conveyed in the delivery of the request, then the likelihood of the request being completed is high. However, like directness and transparency, if the social information is incorrectly assumed or interpreted, then the request becomes much more difficult to comprehend and comply with.

In the examples in Table 1, sentences (2), (3) and (4) are syntactically transparent and pragmatically conventional (Ledbetter & Dent, 1988). All three overtly specify or strongly imply the agent and make the action clear. Thus, in these examples, the request is obvious. In addition to being transparent, these examples are also conventional because they adhere to the sociocultural norms of requesting. Because (4) is syntactically an interrogative, it is considered an indirect request, while (2) and (3) are direct requests because they are imperative sentences.

However, in examples (5) through (8), directness, transparency and/or conventionality are reduced. In (5), while the action is specified, the agent is not. This example is not direct, transparent, or conventional. In (6), the agent is overtly specified, but the agent of the main clause is *I*. The agent is not the intended doer of the action, reducing transparency of the agent and action. Here, the agent is the speaker rather than the listener. Because (6) is phrased as a declarative sentence rather than as an explicit request, it breaks with conventional means for requesting an action and is indirect. In (7), the agent is overtly specified, but the request is phrased in an unconventional manner. This request is indirect and the listener may not interpret the interrogative statement as a request given the mitigating language used. Finally, in (8), the transparency is compromised because the agent is ambiguous (*let's* implies that the speaker

and/or the listener should complete the action). However, conventionality is intact, as this is a polite form of requesting which simultaneously ensures the action is completed without causing unnecessary friction in the social relationship.

The examples in Table 1 are intended to exert the same illocutionary force and achieve the same behavioral outcome, much to the confusion of the listener. Although the examples vary in directness, transparency, and conventionality, they are likely all subject to the same contextual relevance and social information. That is, if the listener can incorporate a shared understanding of contextual demands and social conventions for requesting, then they are more likely to comprehend the request. In this way, contextual relevance and shared understanding of contextual demands may assist in comprehension of indirect requests. However, this may be difficult if the listener is not an adept social communicator or does not understand the contextual demands, as may be the case with young children or individuals with cognitive and linguistic impairments.

Comprehension of Requests

Comprehension of requests for action are traditionally measured by the listener's compliance with the request (Clark, 1979; Ervin-Tripp & Gordon, 1986; Kissine et al., 2015; Kissine et al., 2012; Ledbetter & Dent, 1988). To complete the requested action(s), the listener must comprehend the syntax and vocabulary of the request, correctly identify and interpret the intent of the request, and then finally perform the requested behavior.

Certain request structures may be more difficult to comprehend due to their indirectness, ambiguity, or unconventionality (Clark, 1979; Kissine et al., 2015; Kissine et al., 2012; Ledbetter & Dent, 1988; Ruytenbeek et al., 2017). Indirect requests may be more difficult to comply with because the listener must determine whether the speaker is requesting a literal response or an

indirect response. Returning to example (4), the listener can respond to the literal meaning with "yes, I can," or they can respond to the indirect meaning and perform the action (stir the pudding). In either case, the listener is responding to the request, but in the former, they are incorrectly interpreting the intended meaning of the request. Requests that do not have transparency and/or conventionality, such as (5) through (8), require the listener to identify the intended meaning of the speaker and their goal(s) by determining the requested action and who should perform that action (Clark, 1979). As Clark (1979) explains, this is complicated for the listener. They can provide an expected response, a cooperative but unexpected response, or an uncooperative response. In arriving at the correct interpretation, the listener demonstrates an understanding of the cultural conventions for requesting, the contextual relevance of the request, and the speaker's goal(s).

Development of Request Comprehension

Within the first two years of life, children recognize the word patterns in request structures and rely on gestures paired with words to relate words to objects and actions (Ervin-Tripp & Gordon, 1986). Together, gestures and recognizable patterns allow infants and toddlers to begin comprehending requests. Between 2 and 4 years, children rely heavily on routinized actions such that compliance is engrained in the routine rather than the product of the request type. However, Ervin-Tripp and Gordon (1986) also suggest that children rely on context cues and verbal explicitness rather than suggestions or hints. Thus, at younger ages, children may be more compliant with direct requests. Indeed, this is evidenced in an experiment demonstrating that 3-year-olds are more compliant with direct than indirect requests (Ledbetter & Dent, 1988). However, typically developing 3- and 5-year-olds also demonstrate higher compliance with transparent and conventional indirect requests over ambiguous and unconventional indirect

requests (Ledbetter & Dent, 1988), indicating that young children are compliant with transparent and conventional requests regardless of directness. Therefore, overt specification of the agent and action in combination with the obviousness of request intent are likely important factors in predicting compliance.

Although young children may struggle with interpreting others' goals and points of view, by 7 or 8 years of age, they are aware of the social requirements for use of indirect language in requesting (Ervin-Tripp & Gordon, 1986). Thus, by middle childhood, it is expected that children can comprehend requests for action that vary in directness, transparency, and conventionality. This expectation is borne out in an experiment by Rimac (1985). In his dissertation, Rimac (1985) demonstrated that children with specific language impairment (mean age = 6.46 years) and age-matched (mean age = 6.50 years) and language-matched (mean age = 4.25 years) comparison children complied with over 80% of direct requests and indirect requests. Although this study did not examine transparency and conventionality, it did demonstrate that children with TD are compliant with direct and indirect requests provided in school settings by an unknown examiner by 4 to 6 years of age. It is expected that by 8-years-old, children will be able to comprehend requests of varying directness, transparency, and conventionality (Ervin-Tripp & Gordon, 1986). Based on these findings, adolescents with typical development (TD) should comprehend requests regardless of directness, transparency, and conventionality.

Factors Influencing Comprehension of Requests

Although chronological age and associated cognitive maturation is important for the development of request comprehension, it is not the sole predictor of comprehension and compliance with requests for action. Evidence suggests that language, cognition, and social communication skills may also impact compliance. Several studies have examined the

association between compliance and language ability in children with TD or ASD (Bryce & Jahromi, 2013; Ledbetter & Dent, 1988; Rimac, 1985), but they have reported mixed results. To my knowledge, there have been no investigations into compliance and language ability in children with FXS. Because directness of requests is intertwined with the syntactic structure of the request, it stands to reason that requests which are syntactically more complex may be more difficult to comprehend and comply with than ones that are syntactically simple (Ledbetter & Dent, 1988). In a comparison between 6-year-olds with specific language impairment and age- and language-matched peers, Rimac (1985) reported no difference in compliance to direct or indirect requests based on pragmatic ability and language ability. In contrast, in a study of typically developing 4-year-olds, Ledbetter and Dent (1988) demonstrated that children with higher syntactic comprehension abilities were more compliant overall than those with lower abilities. Specifically, the higher language group was better able to comply with both direct and indirect requests than the lower language group. The lower language group performed better on direct requests with simple syntactic structure than ones with complex syntactic structure. The higher language group performed equally well on the simple and complex direct requests but struggled with the complex indirect requests. This supports the notion that children with higher language ability perform better with more complex or indirect requests than children with lower language ability. However, despite having higher language ability, they are not yet masters of indirect request comprehension at 4-years-old.

In addition to language, nonverbal cognition and the ability to learn may impact compliance with requests in children with and without developmental delays (Ervin-Tripp & Gordon, 1986; Hiebert et al., 2009). In an evaluation of compliance with direct requests, Hiebert et al. (2009) demonstrated that children with and without developmental delays performed in

accordance with their score on an assessment of their "ability to make visual and auditory discriminations" (p. 31). That is, children who scored higher on the test of learning ability had significantly higher likelihood of compliance than those who scored lower, regardless of developmental disability status. Although this demonstrates a link between learning capacity and compliance with direct requests, indirect requests and requests with varying degrees of transparency and conventionality were not assessed. Furthermore, the study by Hiebert et al. (2009) measured capacity for learning, rather than a traditional measure of nonverbal cognition. In contrast to Hiebert and colleagues, nonverbal cognition was not associated with compliance to different types of requests in children with ASD (Kissine et al., 2012). Thus, it remains unclear whether nonverbal cognitive ability is associated with compliance with different request types and whether this association differs between individuals with ASD or FXS.

Extending request comprehension and compliance into ASD, Bryce and Jahromi (2013) reported in a study of 4- to 6-year-old children with high-functioning ASD or TD (n = 20 per group) that both groups were more likely to comply following a direct request than an indirect one. The ASD group was significantly more noncompliant with indirect requests than the TD group. This difference was not accounted for by receptive language, chronological age, or frequency of request type. The authors controlled for receptive language both statistically and through using a language-matched sample. As such, they found no reason to suspect that receptive language impacted compliance in children with high-functioning ASD.

Although receptive language may not have impacted compliance with indirect requests in children with high-functioning ASD, deficits in social communication and pragmatic ability may account for variation in compliance following different types of requests. Requests that are indirect, ambiguous, or unconventional may be more difficult to comply with than those which

are direct, transparent, and conventional, because their intended meanings require pragmatic awareness and adequate social communication skills (Kissine et al., 2015; Kissine et al., 2012). Deficits in social communication are a core feature of ASD (American Psychiatric Association, 2013). As discussed, comprehension of requests lies in shared social information and understanding of the speaker's goal(s) to arrive at the intended meaning of the request. If social communication skills are impaired, such as the ability to infer another person's goals, then comprehension and compliance with indirect, ambiguous, and/or unconventional requests may be compromised. Thus, it is expected that individuals with higher autism symptomology may have difficulty complying with indirect, ambiguous, or unconventional requests.

However, this is not evidenced in studies by Kissine and colleagues (2015; 2012). Kissine et al. (2012) reported on a group of 10 French-speaking children with ASD. They found that compliance was not associated with nonverbal IQ or chronological age. Although they included a measure of language ability, they did not report on associations between language and compliance. Importantly, Kissine et al. (2012) found no difference in compliance by request type – compliance was similarly high for declarative, imperative, sub-sentential (ambiguous) and interrogative requests.

In a follow-up study, Kissine et al. (2015) conducted a semi-structured act-out task which was intended to provide three different scenarios of declarative requesting in a highly controlled context. They found that children with ASD between 7 and 12 years-old were compliant with indirect requests when the request was delivered by an interactional partner, but not when the request was delivered by another speaker with whom the child was not interacting. Thus, they reason that children with ASD are able to comprehend requests that are phrased as declaratives

(e.g., *the pudding needs to be stirred,* which is indirect, ambiguous, and unconventional) when the context demands this interpretation.

Together, these two studies suggest that children with ASD perform well with various requests, regardless of language ability, when the context is contrived and intended to yield a single interpretation of the indirect request. Importantly, these studies fail to consider naturalistic contexts for indirect requesting. Social communication skills are tested when an individual must perform in real-time in an environment that challenges their daily-living skills and preconceived notions of contextual expectations. Additional studies are necessary to examine compliance in naturalistic contexts. Furthermore, these studies do not consider measures of social communication skills and autism symptomology. Moving forward, it will be important to utilize larger samples and to consider skills at all levels while also examining these relationships in varied settings.

Indirect Requesting as a Parenting Strategy

For parents, the use of indirect requests may be purposeful and strategic and may reflect flexible parenting styles. Indeed, appropriate parenting strategies are needed to facilitate compliance in children with challenging behaviors (Bryce & Jahromi, 2013), such as adolescent males with ASD or FXS. Parents may use indirect requests as a means to strategically increase the chances of their child complying. The use of indirect requests provides an illusion of choice for the listener (Clark, 1979), which may be a method for increasing perceived autonomy. Clark (1979) also suggests that indirect requests are used to establish equal authority as opposed to direct requests which may be used to assert control or dominance. These two properties of indirect requests may result in a preference for using indirect requests over direct ones. Additionally, flexible parenting styles may be marked by use of indirect requests. Mothers of

children with Down syndrome (DS) may adjust their requesting behavior based on social contexts and the child's need for supports in a given task (Landry et al., 1994). Young children with DS demonstrate higher compliance when the mother's request follows the child's lead (Landry et al., 1994), suggesting that children's willingness to comply may also be a product of the flexibility of the parent.

Previous research on parenting strategies and parenting styles in FXS suggests that mothers who are overly-directive tend to have children with poorer language and behavior outcomes (Brady et al., 2014; Warren et al., 2010). Mothers who are warmer and more flexible during interactions with their child may promote skill development and language growth which could ultimately foster more independence in their child. Indirect requesting is a form of mitigating language that provides children with choices and opportunities for personal growth and may reflect flexible parenting. However, indirect requests may be difficult for children with language delays and social skill impairments to comprehend (Ledbetter & Dent, 1988). As such, the use of indirect requests with males with FXS or ASD is complicated. If these individuals are unable to comprehend requests that are flexible and promote autonomy, then the use of indirect requests may be ineffective. Thus, it is important to determine the impact of child language on the comprehension of maternal requests and to determine what other individual differences may account for variance in compliance following maternal requests. In doing so, there may be unique strategies that mothers of adolescent males with FXS or ASD can utilize to increase compliance and support their children. Clinical implications may lead to the development of individualized parenting strategies that can maximize outcomes for children with neurodevelopmental disorders. This may provide parents with strategies to balance age- and

language-appropriate requests with increasing opportunities for autonomy while fostering adaptive behaviors and reducing challenging behaviors.

Finally, indirect requests may be particularly relevant during adolescence, when the parent and child are navigating increasing child autonomy. During adolescence, children begin to explore expanding independence and may no longer rely on their parents. As such, parents may change the way they formulate requests for action to resemble how they would pose requests to fellow adults rather than children (Ralph, 2018). By delivering requests as choices or suggestions, the adolescent's independence is increased (Ralph, 2018) and may ensure maximal chances of compliance. By adolescence, it is expected that individuals with TD can comprehend complex requests for actions that vary in directness, transparency, and conventionality. However, this may not be the case for adolescents with developmental delays. Unfortunately, comprehension of requests has not been studied in adolescents with FXS or ASD. Nor has comprehension of requests in adolescents with FXS or ASD been studied in naturalistic settings during mother-child interactions. Accordingly, it will be important to examine parental requesting strategies and their associations with child language ability and compliance. In doing so, requesting strategies used by parents of adolescents with FXS or ASD will be characterized, and the effectiveness of these requests will be identified. Together, these characterizations may inform parenting practices and treatment targets that can be employed to provide individualized parent-child interventions.

Current Study and Research Aims

In three steps, this project examined maternal requesting behavior, adolescent compliance, and adolescent request comprehension during mother-son interactions in adolescent males with FXS and ASD. First, adolescent compliance with maternal requests for action were

considered, along with adolescent-level and maternal request-level predictors of compliance. Next, the impact of autism symptomology and dual diagnosis (i.e., FXS and co-morbid ASD) was assessed as it relates to maternal requesting behavior, adolescent compliance, and adolescent request comprehension during mother-son interactions. Finally, data from a pilot sample of adolescent males with ASD were examined to inform the impact of autism symptomology on maternal requesting behavior, adolescent compliance, and child request comprehension during mother-son interactions. Specifically, the research questions, planned analyses, and hypotheses of each step were as follows.

Aims 1a and 1b: First, does the likelihood of adolescent compliance following a maternal request for action vary as a function of the directness, transparency, and/or conventionality of the request in adolescent males with FXS at either the average or trial-by-trial level? Second, does the likelihood of adolescent compliance following a maternal request for action vary as a function of adolescent language, social skills, or nonverbal cognition?

The first question was examined through correlational analyses with compliance and request properties collapsed across trials (the average level) and through multilevel logistic regression where compliance was predicted by request properties of each individual trial (trial-by-trial level). The second question was examined at the average level (single level logistic regression), as adolescent characteristics did not vary trial-by-trial.

Based on findings by Kissine and colleagues (2015; 2012), it was hypothesized that adolescent compliance would not vary as a function of request properties. That is, request directness, transparency, and conventionality would not predict adolescent compliance at either the average level or the trial-by-trial level. However, it was expected that adolescent language ability, social skills, and nonverbal cognition would predict likelihood of compliance with

maternal requests for action, based on findings in young TD children (Clark, 1979; Ledbetter & Dent, 1988; Rimac, 1985).

Aim 2: Does autism symptomology or FXS+ASD dual diagnosis predict likelihood of adolescent compliance with maternal requests for action? Is a continuous metric of autism symptomology a stronger predictor of likelihood of compliance than a categorical autism diagnosis?

This was tested in several ways. First, based on categorical dual diagnosis, differences in average compliance and adolescent characteristics were described. Then, group membership (+/- ASD) was used to predict average compliance using a single-level logistic regression. Next, a continuous metric of autism symptomology was used to predict average compliance in a single-level logistic regression. Finally, the effect sizes and variance accounted for by each measurement method was compared to determine the relative strength of each predictor.

It was expected that a continuous metric of autism symptomology would better capture individual differences in social communication than a categorical diagnosis and thus would be a stronger predictor of the likelihood of compliance.

Aim 3: The final aim was to preliminarily describe the compliance of adolescent males with idiopathic ASD during mother-son interactions. This step descriptively compared patterns of maternal requests and adolescent compliance in adolescent males with ASD to those of adolescent males with FXS while considering the interplay between autism symptomology, language, and adolescent compliance. It was expected that patterns of compliance would be similar for individuals with ASD as for those with FXS.

Chapter II: Methods

Participants

Children with FXS and their biological mothers were recruited into an ongoing longitudinal study in the FXS Research Lab at the University of Kansas (Brady et al., 2006; Brady et al., 2014; Warren et al., 2010). Participants were recruited from across the United States through a parent listserv, advertising at national conventions, networking with FXS family support groups, and through a national registry at the University of North Carolina- Chapel Hill. Because FXS is a rare disorder, this sample is one of convenience. Although there was some variability in socioeconomic factors, there was limited racial and ethnic variability. Detailed demographic information on the participants with FXS is provided in Table 2.

Thirty-six adolescent males with FXS and their biological mothers contributed data to this project. All adolescents had their FXS diagnosis confirmed through blood testing upon entry into the longitudinal study. Mothers also had their FXS status assessed through blood testing. Of the mothers, 32 carried the premutation (between 55 and 199 CGG repeats), 2 carried the full mutation (200 or more CGG repeats), and 2 had mosaicism for both the premutation and full mutation alleles.

Table 2:

FXS Group Demographic Characteristics

Characteristic	Level	Number	Percentage
Child Ethnicity	Non-Hispanic	33	91.67
	Hispanic	3	8.33
Child Race	White	34	94.44
	Black	4	11.11
	Asian	0	0
	Native American	0	0
	Native Hawaiian or Other Pacific Islander	1	2.78
Mother Ethnicity	Non-Hispanic	33	91.67
	Hispanic	3	8.33
Mother Race	White	34	94.44
	Black	2	5.56
	Asian	0	0
	Native American	0	0
	Native Hawaiian or Other Pacific Islander	1	2.78
Mother Work	No	11	30.56
	Yes	25	69.44
Mother Education	High School Diploma or Equivalent	4	11.11
	Some Post High School	8	22.22
	Associate's or Technical Degree	0	0
	Bachelor's Degree	14	38.89
	Some Post Bachelor's	4	11.11
	Master's Degree	4	11.11
	2 Master's or Doctoral or Professional Degree	2	5.56
Mother Marital Status	Engaged	1	2.78
	Separated	0	0
	Divorced	8	22.22
	Married	25	69.44
	Single, never married	2	5.56
Household Income	> $100,000	24	66.67
	$80,000 - $100,000	2	5.56
	$50,000 - $79,999	6	16.67
	$30,000 - $49,999	0	0
	$15,000 - $29,999	2	5.56
	< $15,000	2	5.56

In addition to the 36 participating families with FXS, two adolescent males with ASD and their mothers were recruited between September 2020 and March 2021. These families were

recruited through advertisements and social media posts in local (Kansas City) organizations, groups and newsletters, teachers and contacts at a school for children with ASD in Colorado, and local community contacts. Demographic information for the ASD group is presented in Table 3, including pseudonyms. All participants provided consent to participate in the study and completed informed consent documents prior to participation.

Table 3:

ASD Group Demographic Characteristics

Characteristic	**Family 1: Casey & Carrie**	**Family 2: Jason & Julie**
Child's Age	15 years	14 years
Mother's Age	51 years	41 years
Child Ethnicity	Non-Hispanic	Non-Hispanic
Child Race	White	White
Mother Ethnicity	Non-Hispanic	Non-Hispanic
Mother Race	White	White
Mother Work	Yes	No
Mother Education	Bachelor's	Some Post Bachelor's
Mother Marital Status	Married	Married
Household Income	$50,000 - $79,999	> $100,000

Procedure

Families with FXS were visited in their homes by two researchers between August 2016 and September 2019. One researcher administered direct assessments of language and cognition with the adolescent and the other completed interviews and parent-reported assessments with the mother. Upon completion of the assessments, the mother and adolescent participated in three 10-minute interactions (doing a puzzle, playing on the iPad, and making a snack). These interactions were video- and audio-taped. After the visit, the two researchers completed an observational measure of autism symptomology, the Childhood Autism Rating Scale – 2nd edition.

Families with ASD met with a researcher through Zoom three times. During the first Zoom meeting, the mother completed the Vineland Adaptive Behavior Scales – II; during the

second, the adolescent completed the Peabody Picture Vocabulary Test – 4th edition; and during the third, the mother and adolescent completed a snack task together. This 10-minute mother-son interaction was video- and audio-recorded. The ASD group did not complete the same battery of measures as the FXS group due to COVID-19 restrictions that were in place at the time of data collection. Specifically, they did not complete the measures of autism symptomology, nonverbal cognition or expressive vocabulary as these required in-person interaction and observation.

Measures

Receptive Vocabulary

The Peabody Picture Vocabulary Test – 4th edition (PPVT-4; Dunn & Dunn, 2007) was used to assess each adolescent's receptive vocabulary. The PPVT-4 is a widely-used standardized assessment in which the participant must identify a picture based on a prompt from the examiner (e.g., "*point to puppy*") from among an array of four pictures. Participants in the FXS group completed the PPVT-4 in person with an examiner during the in-home visits, and participants in the ASD group completed the PPVT-4 over Zoom with the examiner using an electronic version of the assessment. The PPVT-4 yields a raw score (which is equivalent to the total number of items a participant correctly identifies) from which a standard score and an age equivalency can be derived.

Expressive Language

Expressive Vocabulary Test. The Expressive Vocabulary Test – 2nd edition (EVT-2; Williams, 2007) was used to measure expressive vocabulary in the FXS group. This is a widely-used standardized assessment that requires the participant to provide a word based on a prompt from an examiner along with a single image (e.g., *"what's another word for a baby cat?"* along with an image of a kitten). This assessment was administered during the in-home visits by a

trained examiner, and only participants in the FXS group completed this assessment. The EVT-2 yields a raw score which is then converted into a standard score and an age equivalency.

Language Sample. Each adolescent and their mother completed a 10-minute snack task together that served as the context for interaction. From this task, a language sample was collected. Although the length of the task was constant across participants, the number of utterances from each participant varied. The average number of utterances was 94.44, and there was a range from 6 to 213. Number of utterances was included in the analyses as an index of talkativeness. In addition to number of utterances, mean length of utterance in morphemes (MLUm) was calculated as a measure of the adolescent's productive syntax. MLUm was included for all participants, regardless of number of utterances. Work by Casby (2011) has reported no significant differences in MLUm when obtained from 10 or 100 utterances. Utterances were transcribed from the video-recordings using the Noldus Observational Coding system (Noldus Information Technology, 2002) and then analyzed using the Systematic Analysis of Language Transcripts software (SALT; Miller & Iglesias, 2016). Details on transcription and reliability procedures are provided below.

Autism Symptomology

The Childhood Autism Rating Scale (CARS2-ST; Schopler et al., 2010) is an observational and informant-report assessment that was used to measure autism symptomology in the FXS group. The CARS2-ST is a rating scale that consists of 15 items that are each scored along a 7-point scale. Higher scores indicate higher symptom severity. The items describe various behaviors, including verbal and nonverbal language skills, adaptation to change, emotional response, unusual sensory interest, abnormal body use, and self-injurious behaviors. When completing this assessment, the examiner can consider multiple sources of information

including in-person observation and interaction, parent or informant reporting, and other testing, such as cognitive testing. The total score on the CARS2-ST is the sum of the 15 items. For individuals over 13 years-old, scores below 28 suggest *few-to-no symptoms of autism*, scores higher than or equal to 28 are classified as *mild-to-moderate symptoms of autism*, and scores greater than or equal to 34 indicate *severe symptoms of autism*. The cut-offs for each category are slightly different for individuals younger than 13 years-old: < 30, ≥ 30, and ≥ 37, respectively.

Nonverbal Cognition

The Leiter International Performance Scale – Revised (Leiter-R; Roid & Miller, 1997) was used to assess nonverbal cognition in the FXS group. This assessment is administered in person, and during the examination, the examiner cannot use verbal communication. Participants completed the Brief IQ test which is formed of four subtests: figure ground, form completion, repeated patterns, and sequential ordering. The Brief IQ test yields an IQ score, an age equivalency, and a growth scale value which are derived from the raw score. Growth scale value was used in the analyses since it is an index of true ability (Farmer et al., 2020) and because of the strong floor effects for the IQ score.

Social Skills

The Vineland Adaptive Behavior Scales, second edition (VABS-II; Sparrow et al., 2005) was used to assess social skills in the FXS and ASD groups. Mothers completed this interview during the home visits (FXS group) or through Zoom (ASD group), which took 45 to 60 minutes. This parent interview assesses adaptive behavior in three domains (communication, social skills, and daily-living skills) and also includes four maladaptive behavior subscales (internalizing, externalizing, critical, and other). Although the entire interview was administered, only the social

skills domain was included in this project. Each item in the assessment is scored along a three-point scale, with higher scores indicating higher ability.

Mother-Son Interactions

Adolescents and their mothers completed a 10-minute snack task together in their homes. They were provided with the materials to make dirt cups (chocolate pudding with Oreos and gummy worms) and instructed that they should spend 10 minutes making the snack together. The task was recorded on a video camera and an audio recorder for later transcription. Using the Noldus Observational Coding system (Noldus Information Technology, 2002), all mother and adolescent behaviors and communication during the interactions were transcribed and coded.

Transcription. Every communication turn from the mother and adolescent was transcribed in Noldus, then converted into a text document, and then analyzed in SALT (Miller & Iglesias, 2016). Mother and adolescent turns were segmented into utterances by C-units, which were defined as main clauses with all subordinate clauses. Thus, a turn could include multiple C-units (or utterances). Each utterance was also segmented by bound morphemes in accordance with the SALT manual which contributed to MLUm scoring.

Coding. Mother and adolescent behaviors were coded from the snack task videos. Relevant to this project, mother communication was coded based on function of the communication act. This included requests for verbal compliance, comments, recodes, and requests for behavioral compliance, referred to in this manuscript as requests for action. Each request was then coded for request properties and labeled as direct or indirect, transparent or ambiguous, and conventional or unconventional. Request property coding was based on the transcript, rather than on the videotapes. Specific definitions and the coding manuals are included in the Appendix.

Adolescent behaviors and communication were also coded by morphology and function, respectively. Of importance to this project is adolescent compliance which was coded in response to each maternal request for action. Adolescent compliance was coded as either *compliant*, *noncompliant*, or *no opportunity*. A *compliant* action occurred when the adolescent completed the mother's request, either in full or in part. A *noncompliant* action occurred when the adolescent did not attempt to complete the mother's request. A *no opportunity* occurred when the mother completed the action herself, physically assisted the adolescent in completing the action, or when she did not provide the adolescent with sufficient time to complete the action before delivering another request. If the mother gave multiple requests in the same turn, (e.g., "Put the pudding in the jar. Put it in the jar. Open it up all the way.") the adolescent's compliance with the last request was considered. All instances of maternal requests for action followed by *no opportunity* compliance codes were discarded and not considered in the current analysis. The coding manuals for maternal requests for action, request properties, and adolescent compliance are included in the Appendix.

Reliability. Reliability for the Noldus transcription and behavioral coding was through consensus coding. For each file, a coder independently transcribed and coded the mother's communication and the adolescent's communication and behaviors. After the first coder finished, a second coder then reviewed the transcripts and codes independently from the first coder. The second coder noted any disagreements with transcription and coding and then the two coders reviewed the transcripts and codes together. All disagreements were resolved through consensus. Thus, each file was reviewed three times: once by the original coder, once by the second coder, and then once during a consensus coding meeting.

Request property coding took place separately from the Noldus coding (i.e., the maternal request coding). This variable was unique to this project so it was not included in the original Noldus coding. Reliability for the request property coding utilized primary and secondary coders. Coders were trained to 90% agreement across three files before coding independently. One third of files (33%) were coded by both the primary and secondary coders. Percent agreement and Kappa values were calculated as indices of reliability. For directness, percent agreement was 91.44% and K = .80; for transparency, 93.39% and K = .84; and for conventionality 95% and K = .85.

Chapter III: Results

Analysis Plan

Data were analyzed through several different levels of analysis, depending on the research aim. Adolescent characteristics such as language ability, social skills, and average level of compliance were characterized descriptively, as were maternal requesting behaviors. Correlations between variables were assessed to examine the strength and direction of the relationship between adolescent abilities, maternal requesting, and adolescent compliance. The first two types of analysis were conducted using IBM SPSS for Windows, version 25 (IBM Corp., 2017). Then, multilevel logistic regression modeling was used to assess the extent to which maternal requesting and adolescent characteristics predicted adolescent compliance. Analyses utilized full-information marginal maximum likelihood estimation based on LaPlace approximation in SAS software version 9.4 with PROC GLIMMIX (SAS Institute Inc., 2013). Multilevel modeling is a form of regression analysis that allows for examination of change over time while considering the impact of time-varying and time-invariant predictors.

In the analyses, raw scores for the PPVT-4, EVT-2 and VABS-II were used rather than standard scores. Similarly, growth scale value scores were used for the Leiter-R rather than the derived IQ score. The use of standard scores to document individual differences has recently come into question (Farmer et al., 2020), as standard scores may not accurately reflect ability in individuals who progress at slower than expected rates, such as those with FXS or ASD. As such, raw scores or growth scale values on these standardized measures may better reflect the variation in ability among this sample of adolescents. Indeed, 44% of adolescents in the FXS group had standard scores on the PPVT-4 at the floor, suggesting a strong floor effect in this sample. Similarly, over 65% of Leiter-R IQ scores were at the floor. Thus, raw scores were used

for the PPVT-4, EVT-2, and VABS-II. Raw scores were chosen in lieu of growth scores for the PPVT-4 and EVT-2 because they are more readily interpretable. Raw scores were used on the VABS-II because the VABS-II does not provide growth scale values; rather it provides age equivalencies. Finally, total raw scores were used for the CARS2-ST.

Descriptives

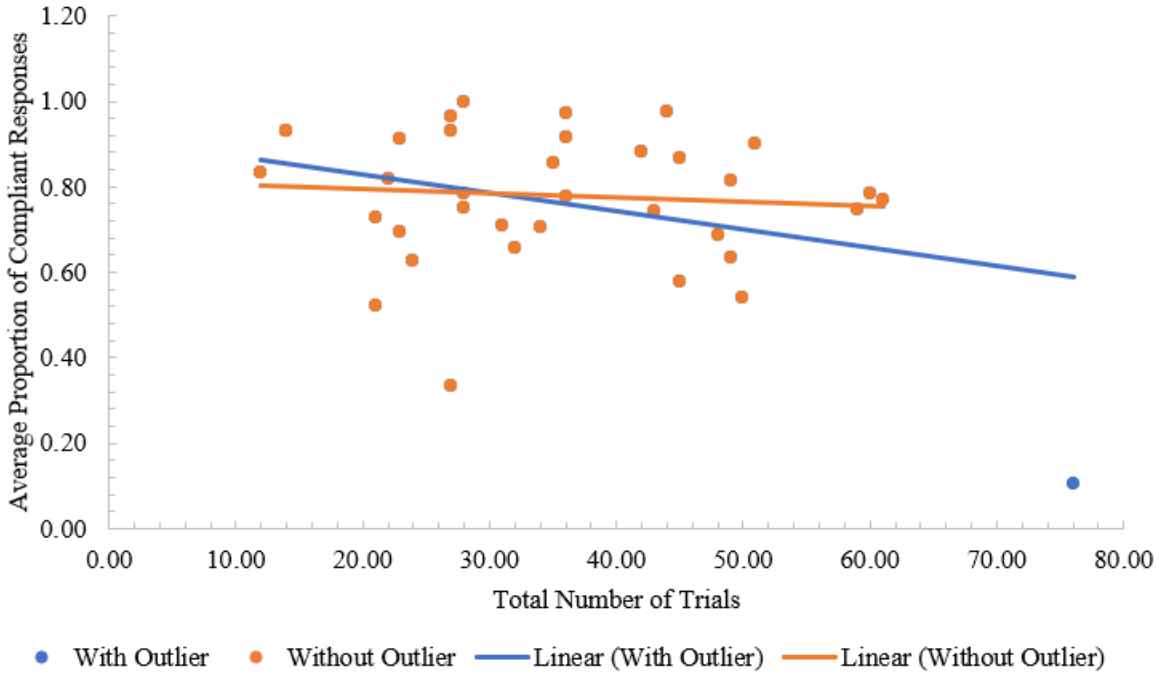
The adolescents with FXS ranged in age from 12.42 to 18 years, with an average age of 16.13 years (+/- 1.08). As shown in Table 4, they varied in language ability, with PPVT-4 raw scores ranging from 24 to 147, EVT raw scores ranging from 6 to 114, MLUm ranging from 0 to 3.53, and number of utterances ranging from 6 to 213. Variability in raw scores on the VABS-II indicated individual variation in social skills. Nonverbal cognition was also variable, although all adolescents had nonverbal IQs greater than 3 standard deviations below the normative mean (100), suggesting all adolescents had moderate intellectual disability.

There was variability in the total number of maternal requests for action. The total number of requests for action, also referred to as the number of trials, was used as the index of time for the multilevel modeling. The total number of requests ranged from 12, or a rate of 1.2 per minute, to 76, or a rate of 7.6 per minute. On average, mothers utilized requests for action 3.6 times per minute, or 36 times during the 10-minute task. The standard deviation of number of requests was 14.83, suggesting considerable variability between mothers. Eighty percent of mothers used between 21 and 51 requests, suggesting that the few who used less than 21 (n = 3) or greater than 51 (n = 4) may be outlying cases. Upon closer examination, it was found that one case was outlying in both person-level average proportion of compliant responses and in total number of requests for action, as shown in Figure 1. This outlying case was not included in the subsequent correlations or multilevel modeling. However, the descriptive statistics and

demographic information include the outlier to provide a representative description of the variability seen in males with FXS.

Figure 1:

Association Between Number of Requests for Action and Within-Person Average Compliance



Eight adolescents were minimally verbal and had fewer than five different words in the 10-minute language sample from the mother-child interaction. These eight adolescents primarily communicated using gestures and signs, and one used a speech-generating device in combination with gestures and signs. Seven of these individuals were unable to complete the practice items on the EVT-2, and thus the EVT-2 was not administered to these participants. One was unable to complete the PPVT-4 and three were unable to complete the Leiter-R due to challenging behaviors during the home-visit.

Table 4:

FXS Group Descriptive Statistics

	N	Min	Max	Mean	SD
Age in Months	36	149	216	193.58	12.91
Age in Years	36	12.42	18.00	16.13	1.08
PPVT-4 Raw Score	35	24	147	81.14	39.13
EVT-2 Raw Score	29	6	114	68.66	28.44
MLU in Morphemes (MLUm)	36	0	3.53	2.16	1.01
Number of Utterances	36	6	213	94.44	64.06
Leiter-R IQ	33	36	52	37.82	3.69
Leiter-R Growth Scale Value	33	404	474	452.15	15.62
VABS-II Social Skills Raw Score	36	35	188	118.81	45.33
CARS2-ST Total	36	17.50	39.00	25.97	6.55
Total # Trials	36	12	76	36.14	14.83
Proportion of Compliant Actions	36	.11	1.00	.76	.19
Proportion of Direct Requests	36	.32	.91	.67	.13
Proportion of Transparent Requests	36	.47	.96	.74	.11
Proportion of Conventional Requests	36	.45	.96	.78	.11

For each mother, the proportions of direct, transparent, and conventional requests were calculated as:

$$Prop.Direct = Total\ \#\ Direct\ Requests/Total\ \#\ Requests$$

Similarly, for each adolescent, the proportion of compliant responses to the maternal requests for action was calculated as:

$$Prop.Compliant = Total\ \#\ Compliant\ Responses/Total\ \#\ Responses$$

On average, one-third (67%) of maternal requests for action were direct requests, suggesting mothers used a higher proportion of direct than indirect requests. However, this varied between mothers, as the range was 32% to 91%, see Table 4. Transparency was more frequent than directness, as was conventionality. On average, mothers used transparent and conventional requests 74% and 78% of the time, respectively. As with directness, there was considerable variability in transparency and conventionality. Adolescents were largely compliant with maternal requests for action, with an average level of compliance equal to 78%. Again, however,

there was considerable variability between persons, as the range in compliance was between 33% and 100%.

Correlations

Correlations were examined to determine the association between proportion of compliant responses and proportion of direct, transparent, and conventional requests and adolescent characteristics. This method of analysis is a broad approach to determine basic associations between variables but does not provide trial-by-trial predictive information. As such, the correlational analyses here inform the association between average compliance and other variables but do not predict a categorical compliant response.

Proportion of compliant responses was correlated with all language measures such that higher compliance was correlated with higher language ability. Compliance was also significantly positively correlated with social skills and significantly negatively correlated with autism symptoms. Similarly, proportion of compliant actions was significantly associated with nonverbal cognition such that higher compliance was associated with higher nonverbal cognitive ability. The proportion of direct requests was negatively correlated with the proportion of compliant actions; however, this association was not significant ($p = 0.09$), see Table 5. Average transparency and conventionality were not associated with average compliance, nor with any adolescent characteristics.

Table 5:

Correlations with Proportion of Direct Requests and Proportion of Compliant Actions

	Prop. Direct	**Prop. Transparent**	**Prop. Conventional**	**Prop. Compliant**
			r (*p*-value)	
Prop. Compliant	-.29 (.09)	.01 (.96)	-.09 (.61)	
PPVT-4 Raw Score	.02 (.91)	.16 (.38)	.09 (.62)	.56 (.001)
EVT-2 Raw Score	-.12 (.54)	.08 (.70)	.12 (.54)	.53 (.004)
MLUm	.08 (.66)	.12 (.51)	-.05 (.80)	.52 (.002)
Number of Utterances	.08 (.65)	.12 (.48)	.14 (.43)	.27 (.012)
Leiter-R Growth Scale Value	.14 (.45)	.13 (.47)	.08 (.67)	.53 (.002)
VABS-II Social Skills Raw Score	.06 (.72)	.12 (.51)	.09 (.61)	.46 (.01)
CARS2-ST Total	-.13 (.44)	-.27 (.12)	-.26 (.13)	-.40 (.02)

Because the language variables were significantly and strongly correlated with one another, see Table 6, they were reduced to a single composite score that encompassed expressive language, receptive language, and nonverbal cognition. Leiter-R growth scale values were included in the composite language score because numerous studies have demonstrated that nonverbal cognition is strongly associated with language and accounts for significant variance in language in FXS (McDuffie et al., 2012; Roberts et al., 2001; Roberts et al., 2007). Indeed, the Leiter-R was strongly correlated with the PPVT-4, the EVT-2, and MLUm (all *r*s > .60, *p*s < .01). Each of the language and nonverbal cognition measures were transformed into z-scores to standardize the scale of the scores which ensures that variance is equal across variables (Song et al., 2013). The z-scores were then averaged together to create the composite language score for each adolescent. Averaging was chosen over a summative score because of missing data on the EVT-2 and Leiter-R. The new composite language score was very strongly correlated with each of the variables from which it was derived, demonstrating that the composite language score was

an adequate substitute for the assessment scores, see Table 6. Finally, because the composite language score was the mean of the z-scores, it could be interpreted that a score of 0 represents the mean performance.

Table 6:

Correlations Between Predictors and Composite Language Scores

	Mean	SD	n	1	2	3	4	5	6	7
1. VABS-II Social Skills Raw	118.20	45.84	35	-						
2. CARS2-ST Total	25.73	6.48	35	-.82**	-					
3. PPVT-4 Raw	81.41	39.69	34	.66**	-.72**	-				
4. EVT-2 Raw	68.75	28.95	28	.53**	-.74**	.92**	-			
5. Leiter-R Growth Scale Value	452.16	15.87	32	.63**	-.72**	.80**	.74**	-		
6. MLU in Morphemes	2.16	1.01	35	.78**	-.77**	.77**	.78**	.64**	-	
7. Number of Utterances	94.44	64.06	36	.60**	-.69**	.49**	.32	.36*	.69**	-
8. Composite Language Score	-.22	.78	36	.77**	-.85**	.91**	.91**	.82**	.91**	.74**

Note: ** $p \leq .01$; * $.01 > p \leq .05$

Aim 1a: Likelihood of Compliance as a Function of Maternal Request Properties

Multilevel modeling was used to examine the predictive abilities of maternal requesting properties on adolescent compliance. This method of analysis enables trial-level prediction of a categorical compliant response, which is a fine-grained approach to assessing what might predict probability of adolescent compliant actions to maternal requests.

Step 1: Empty Model

The extent to which number of trials and request properties predicted adolescent compliance was assessed through a series of multilevel models in which the 1,226 trials were modeled as nested at Level-1 within the 35 dyads at Level-2. Differences between dyads were assessed through person-level random effects. The binary compliance outcome was predicted

using a logit link function and binary conditional outcome distribution. All model parameters were estimated via full-information marginal maximum likelihood using LaPlace integral approximation in SAS PROC GLIMMIX. The significance of fixed effects was evaluated with Wald tests, which are the *t*-test of the ratio of each estimate to its standard error using between-within denominator degrees of freedom. Significance of random effects was evaluated via likelihood ratio tests, which are the -2ΔLL with degrees of freedom equal to the number of new random effects variances and covariances. Effect sizes for the fixed effects were evaluated via odds ratio estimates which indicate the odds of a compliant response.

To begin, an empty means, random intercept model was estimated to determine the proportion of variance in probability of compliance that was due to Level-1 and Level-2 sources. This model indicated that 17.3% of the variance in compliance was between dyads, -2LLΔ (1) = 58.38, p <0.0001, suggesting significant variation in probability of compliance between adolescents. A 95% random effects confidence interval, calculated as fixed intercept ±1.96*SQRT(random intercept variance), indicated that 95% of the adolescents were predicted to have intercepts for probability of compliance between .45 and .95. The average estimate for the fixed intercept for the log-odds (logit) of complying was 1.40, or a probability of .80.

Step 2: Effect of Number of Trials

Next, the impact of number of trials in predicting probability of compliance was examined. A higher number of trials indicates a higher rate of requests per minute during the mother-son interaction. This was assessed before examining request properties because it is important to account for the effect of time prior to adding other predictors. A fixed effect of linear trial was assessed and results are shown in Table 7. The effect of linear trial (slope) in logits was equal to -0.007, p = .19, suggesting there was no significant effect of trial on

compliance. The addition of a random effect of trial slope did not improve model fit, -2LLΔ (2) = 4.27, p = .08. The effect of number of trials was not included in subsequent analyses.

Table 7:

Multilevel Models for Number of Trials and Request Properties

Model Effects	Step 1: Empty Means, Random Intercept		Step 2: Trial		Step 3a: Directness		Step 3b: Transparency		Step 3c: Conventionality	
	Est.	SE	Est.	SE	Est.	SE	Est.	SE	Est.	SE
Model for the Means										
Intercept	1.40**	0.16	1.54**	0.20	2.44**	0.79	1.15	1.11	1.85	1.21
Number of Trials			-0.007	0.006						
Request Property Effects										
Level-2					-1.57	1.15	0.33	1.47	-0.57	1.53
Level-1					0.14	0.16	-0.15	0.17	-0.12	0.18
Model for the Variance										
Random Intercept Variance	0.67**	0.24	0.67**	0.24	0.60**	0.22	0.67**	0.24		
-2 Log Likelihood	1259.12		1257.44		1256.59		1258.23		1258.55	
AIC	1263.12		1263.44		1264.59		1266.23		1266.55	
BIC	1266.23		1268.11		1270.81		1272.45		1272.77	

Note: ** $p \leq .01$; * $.01 > p \leq .05$

Step 3: Effect of Request Properties

Next, in line with the research questions, directness, transparency, and conventionality of maternal requests for action were examined as potential predictors of adolescent compliance, see Table 7. First, directness was added to the model at level-1 and level-2. Person-mean-centering was used to partition directness into level-1 and level-2 effects. Directness at level-1 represents the trial-varying binary directness (1 = direct, 0 = indirect) of each individual maternal request for action. Level-2 directness represents the trial-invariant average directness for each mother (e.g., if the mother gave 20 requests in total and 18 were direct, then her level-2 directness would

be 18/20 = .90). Request directness at level-1 and level-2 did not significantly predict child compliance, $t = 0.86$, $p = .39$ and $t = -1.36$, $p = .18$, respectively. Although request directness did not predict compliance, the effect size for level-1 directness was at 1.15 (95% CI: 0.84-1.58). The effect size for level-2 directness was very small at 0.21 (95% CI: 0.02-2.17).

Next, transparency and conventionality were added to the model, individually. Like directness, transparency was not predictive of compliance at level-1 or level-2, $t = -0.92$, $p = .36$ and $t = 0.22$, $p = .82$, respectively. The effect size for level-1 transparency was 0.86 (95% CI: 0.62-1.19). The effect size for level-2 transparency was 1.39 (95% CI: 0.06-27.93). Finally, conventionality also did not predict adolescent compliance at level-1 or level-2, $t = -0.66$, $p = .51$ and $t = -0.37$, $p = .71$, respectively. The effect size for level-1 conventionality was 0.89 (95% CI: 0.63-1.26) and for level-2 conventionality was 0.56 (95% CI: 0.03-12.64). Request properties did not seem to impact the likelihood of an adolescent complying with requests at the trial-by-trial level nor across trials (i.e., average level). Importantly, standard error estimates were quite high for level-2 transparency and conventionality, suggesting the sample may have been too small or that there were other factors affecting compliance that were not controlled.

Aim 1b: Likelihood of Compliance as a Function of Adolescent Characteristics

The effects of adolescent language and social skills were assessed next. Because adolescent characteristics did not vary by trial (i.e. they were measured only once, or cross-sectionally), these predictors were considered as trial-invariant and thus only accounted for variation at level-2 (the between-person level). As such, these predictors did not need to be partitioned into level-1 and level-2 effects. Adolescent language was represented by the composite language score as described above.

Alone, adolescent language was significantly predictive of compliance, $t = 3.87$, $p < .001$, such that higher language ability predicted higher likelihood of compliance. The intercept for adolescent language represented an individual with average language skill relative to the rest of the sample. So, for each unit increase in adolescent language, the predicted increase of log odds (logit) of compliance was 0.64. This translated to an effect size for adolescent language, calculated as an odds ratio (OR), of 1.91 (95% confidence interval: 1.36 – 2.67). This OR indicates that the expected multiplicative increase in probability of compliance given a one unit increase in composite adolescent language ability is 1.91 times greater. The effect of adolescent language accounted for 39.09% of the between-person random intercept variance.

When adolescent social skills, as measured by the VABS-II social skills domain raw score, were added, the fixed effect of social skills was not significant, t = -0.07, p = .94 while the effect of adolescent language remained significant, see Table 8. The effect size for social skills was equal to 1.00 (95% confidence interval = 0.99 to 1.01). Although social skills were not a significant predictor, they were significantly correlated with proportion of compliant responses, suggesting that higher social skills are associated with higher average compliance. The model with language and social skills did not account for additional variance as compared to the language-only model. Table 8 provides estimates and standard errors for both models.

Table 8:

Multilevel Models for Adolescent Characteristics

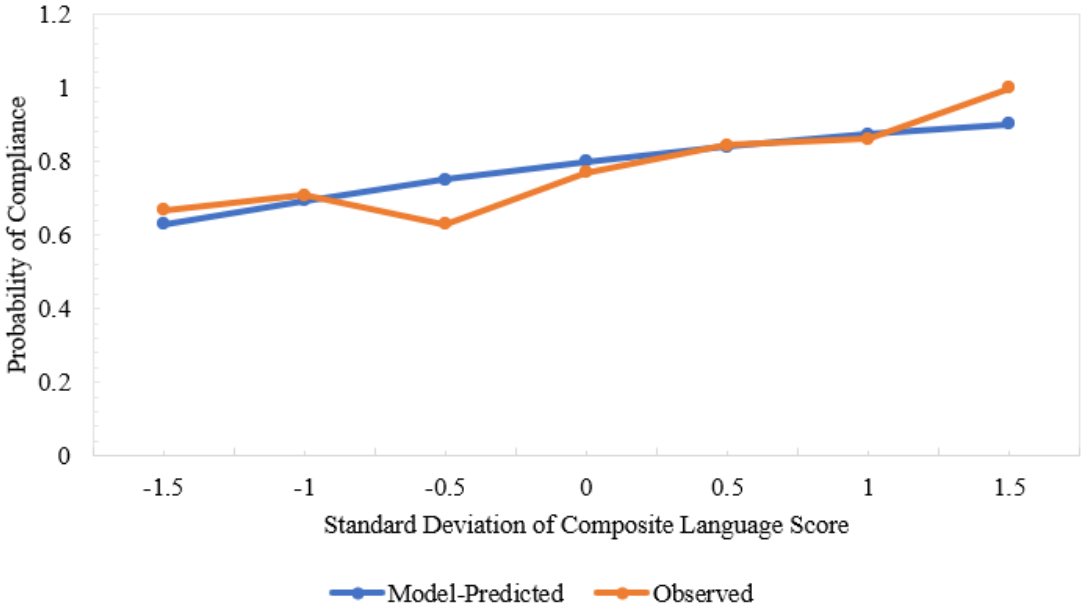
Model Effects	Step 1: Language		Step 2: Add Social	
	Est.	SE	Est.	SE
Model for the Means				
Intercept	1.57**	0.15	1.61*	.63
Time-invariant Predictors				
Composite Language Ability	0.64**	0.17	0.66*	0.26
VABS-II Social Skills Raw Score			-0.0003	0.005
Model for the Variance				
Random Intercept Variance	0.41**	0.16	0.42**	0.17
-2 Log Likelihood	1245.56		1240.55	
AIC	1251.56		1248.55	
BIC	1256.23		1254.77	

Note: ** $p \leq .01$; * $.01 > p \leq .05$

Figure 2 depicts the model predictions for probability of compliance based on the most parsimonious model (composite language score only). Observed probability of compliance given certain composite language scores are shown along with the model-predicted probability of compliance. Along the x-axis is the mean composite language score (0) and then increasing and decreasing scores based on half standard deviations. The predicted probability of compliance for an individual with mean adolescent language was .80 and the observed probability of compliance for an individual with mean adolescent language was .77. Both the observed and predicted probabilities of compliance increased as adolescent language increased. According to the model, an adolescent with language ability one standard deviation below the mean would have probability of compliance of .70 and an adolescent whose language is one standard deviation above the mean would have a probability of compliance of .87.

Figure 2:

Observed and Model-Predicted Compliance by Composite Language Score



Aim 2: Autism Symptoms/Diagnosis and Likelihood of Compliance in FXS

The next set of analyses were aimed at examining the impact of autism symptomology and co-morbid FXS and ASD (referred to as FXS+ASD) on the likelihood of compliance following a maternal request for action. Autism symptomology was measured as a continuous predictor by the total score on the CARS2-ST. Scores ranged from 17.5 to 39, with a mean of 25.73 (+/- 6.48). When autism symptomology was tested as a predictor of adolescent compliance in the multilevel model, it was significantly predictive of likelihood of compliance, t = -2.52, p = .02. For every point higher on the CARS2-ST, the logit for the probability of complying to a maternal request for action was significantly lower by 0.058, which translates to an OR of 0.94 (95% CI: 0.90-0.99). This between-person effect accounted for 19.79% of the level-2 person random intercept variance.

Co-morbid FXS and ASD was defined based on a cut-off on the CARS2-ST. Two groups were created, one where adolescents had CARS2-ST total scores greater than or equal to 27.5 (the FXS+ASD group), per the publisher's specifications, and one where adolescent's scores were less than 27.5 (the FXS-O group). There were 14 participants in the FXS+ASD group and 21 in the FXS-O group. Differences between the means for select variables were analyzed through ANOVAs and are presented in Table 9. Groups differed significantly on all language and social skill measures, with the FXS-O group performing higher than the FXS+ASD group. Groups did not differ on age, total number of maternal requests for action, nor maternal request properties. However, groups differed significantly on compliance, with the FXS-O group having significantly higher average compliance than the FXS+ASD group.

Table 9:

FXS-O and FXS+ASD Between-group Differences

Variable	**FXS-O Mean (SD)**	**FXS+ASD Mean (SD)**	**F**	***p*-value**
Age in Years	16.23 (0.87)	15.99 (1.38)	0.39	.54
Composite Language Score	0.26 (0.54)	-1.03 (0.56)	46.17	< .001
PPVT-4 Raw Score	104.15 (27.68)	48.93 (30.76)	29.92	< .001
EVT-2 Raw Score	80.96 (19.67)	38.25 (26.36)	22.17	< .001
MLU in morphemes (MLUm)	2.73 (0.63)	1.30 (0.84)	33.29	< .001
Number of Utterances	128.10 (50.12)	41.43 (47.23)	26.28	< .001
Leiter-R Growth Scale Value	460.05 (8.99)	439.00 (16.38)	22.23	< .001
VABS-II Social Skills Raw Score	147.14 (29.92)	74.79 (27.17)	52.79	< .001
Total # Trials	36.57 (14.73)	32.64 (11.06)	0.72	.40
Proportion of Compliant Actions	.83 (.11)	.71 (.18)	6.37	.017
Proportion of Direct Requests	.68 (.12)	.65 (.16)	0.30	.59
Proportion of Transparent Requests	.77 (.10)	.71 (.12)	2.23	.15
Proportion of Conventional Requests	.80 (.09)	.75 (.13)	1.86	.18

Examining autism diagnostic category in the regression model, adolescents in the FXS-O group were more likely to comply than adolescents in the FXS+ASD group. Relative to the FXS+ASD group, the logit for the probability of complying in the FXS-O group was

significantly higher by 0.76 (t = 2.53, p = .02), which translates to an OR of 2.13 (95% CI: 1.16-3.91). The fixed effect of categorical autism status accounted for 21.2% of the level-2 random intercept variance.

In both regression models, adolescents with higher autism symptomology were less likely to comply than adolescents with lower autism symptomology. However, the OR, which is a measure of effect size, was larger in the categorical model than the continuous model. That is, ASD group was a stronger predictor than autism symptom severity based on effect size. Similarly, the variance accounted for by autism diagnostic category was slightly higher than that of autism symptom severity.

Aim 3: Preliminary Findings in an ASD Group

Two adolescent males, Casey and Jason (all names changed to protect identities) with idiopathic ASD and their mothers, Carrie and Julie, provided data through Zoom. The adolescents both completed the PPVT-4 online and their mothers completed the VABS-II. Then, the dyads completed the 10-minute snack task. Demographic information is provided in Table 3 and scores on the assessments are provided in Table 10. Casey and Jason were both low verbal communicators, meaning they did not have large expressive vocabularies. Casey had a difficult time completing the PPVT-4, as he was largely noncompliant with the assessor's directions and did not enjoy working over Zoom. Thus, his PPVT-4 raw and standard scores reflect a floor effect. Jason was more compliant with the assessor over Zoom and his scores reflect his engagement and his ability. Both communicated using signs, gestures, vocalizations, and verbalizations. The language and social scores for Casey were low but fell within the range of scores of the FXS group on all but the PPVT-4 raw score. Jason's scores were all within the range of the FXS group and his PPVT-4 scores were above the FXS group average.

Table 10:

Assessment and Task Scores for Casey and Jason

Score	Casey	Jason	FXS Mean (Range)
PPVT-4 Raw Score	4	98	81.40 (24 – 147)
PPVT-4 Standard Score	20	49	38 (20-71)
VABS-II Social Skills Raw Score	42	60	118.2 (35-188)
# of Utterances	18	40	94.44 (6-213)
MLUm	0.17	0.65	2.16 (0-3.53)

Both families received their snack-materials in the mail along with instructions for how to complete the task. The total number of requests for action was commensurate with the FXS group, as were the proportions of compliant responses, direct requests, transparent requests, and conventional requests, see Table 11. Neither dyad appeared to be an outlier. As with the FXS group, Casey and Jason were largely compliant and their mothers tended to use direct, transparent, and conventional requests. However, with the limited data in the ASD group, statistical tests are underpowered and should be interpreted with caution. Figures 3 and 4 show the relative frequency of compliance given different request properties for the ASD and FXS groups, respectively. Both groups demonstrate a similar pattern of compliance, with the most compliance occurring with direct, transparent, or conventional requests. For example, in the FXS group, when examining compliance and the request property of directness, the combination [+ compliant, + direct] is the most frequent of all compliant and direct combinations. That is, the adolescents are most often compliant when the request is direct as opposed to when it is indirect. This pattern was true for all request properties and across groups. The preliminary data from two participants with ASD so far suggests similar trends as the larger FXS group.

Table 11:

ASD Group Requests and Compliance

Variable	Casey	Jason	FXS Mean (Range)
Proportion Compliant	.63	.93	.78 (.33-1)
Proportion Direct	.67	.63	.67 (.32-.91)
Proportion Transparent	.93	.88	.74 (.47-.96)
Proportion Conventional	.87	.93	.78 (.45-.96)
# of Requests	30	41	35 (12-61)

Figure 3:

ASD Group Relative Frequency of Compliance by Request Properties

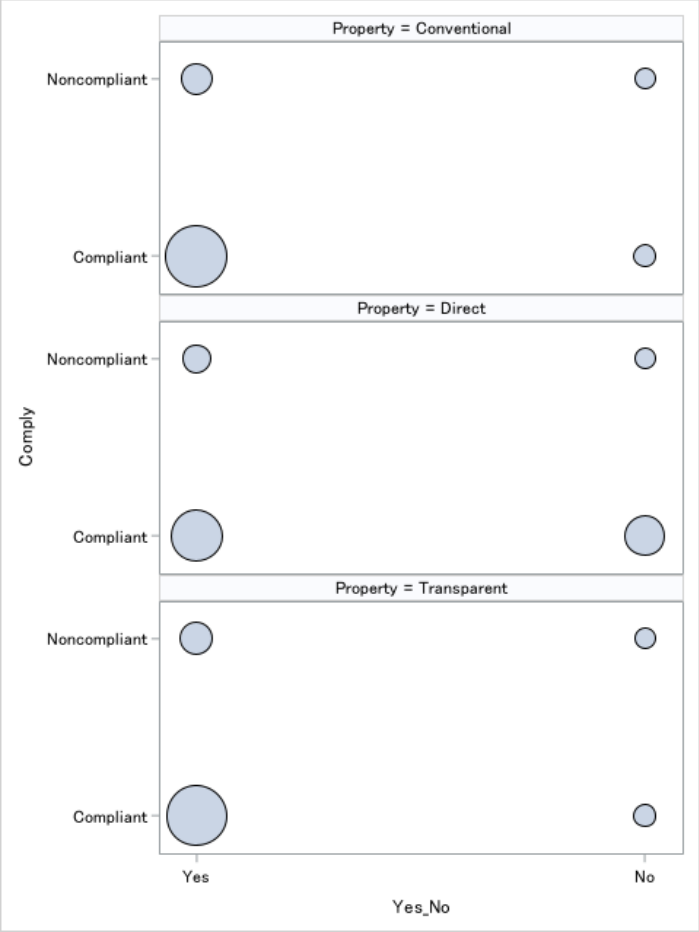
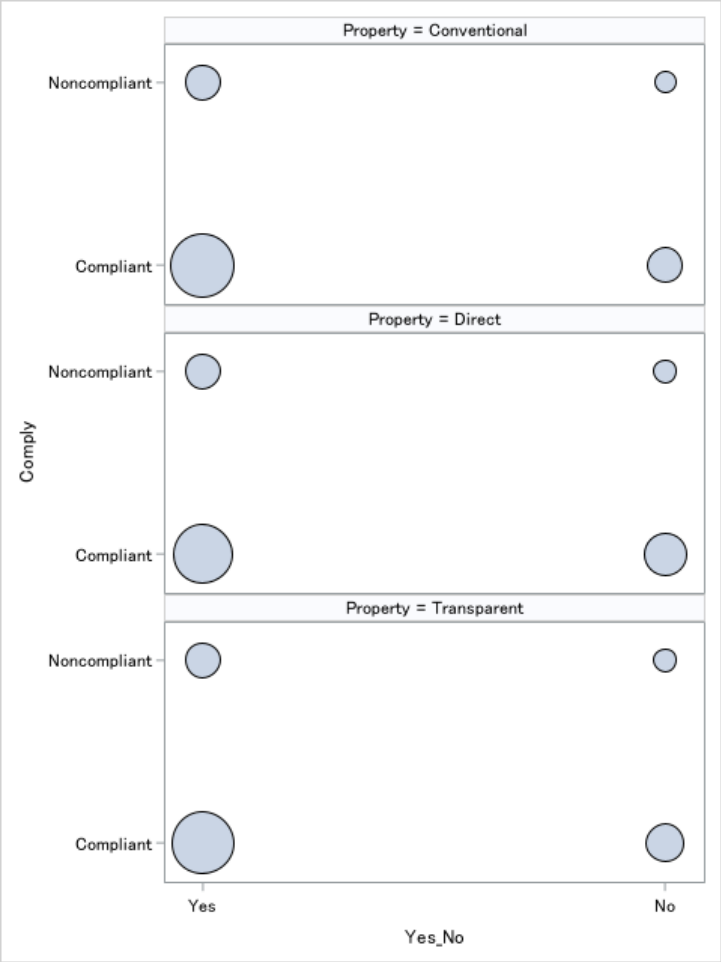


Figure 4:

FXS Group Relative Frequency of Compliance by Request Properties



Given that there were only two participants with ASD, correlational analyses between language and compliance were not practical. However, it is relevant to note that Casey had lower compliance and lower language ability (both receptively and based on the 10-minute mother-child interaction) while Jason had higher compliance and higher language ability. This pattern was seen in the FXS group and is supported in the ASD group, but more data is necessary to make any comparisons or draw any meaningful conclusions. Similarly, because there were only two participants in this group, it was not possible to assess the interplay between autism symptoms and compliance. Casey and Jason both had confirmed ASD diagnoses, and since data

was collected over Zoom a formal assessment of autism symptomology, such as the CARS2-ST or the Autism Diagnostic Observation Schedule (Lord et al., 2012), was not completed.

Chapter IV: Discussion

Across adolescents, compliance was high. Within the combined FXS group, adolescents complied with 76% of requests on average and the two adolescents with ASD complied with 63% and 93% of requests, respectively. These findings are in line with those of Kissine et al. (2012) who observed relatively high levels of compliance in a study of children with ASD. When prompting for compliance, mothers tended to use direct, transparent, and conventional requests more often than indirect, ambiguous, or unconventional ones. The results did not suggest that adolescents were more compliant with one request type over another. Additionally, there was no effect of trial, indicating that adolescents were just as likely to comply later in the session than earlier. While none of the trial-level predictors were significantly predictive of compliance, adolescent-level predictors (i.e., language, nonverbal cognition, social skills, and autism symptomology) were significantly predictive.

Maternal Requesting Behaviors

Maternal requesting behaviors have been shown to vary across disability groups and contexts (Bryce & Jahromi, 2013; Landry et al., 1994). In their study, Landry and colleagues (1994) found that mothers of young TD children used a higher proportion of suggestive requests (e.g., “where does this one go?”) than mothers of young children with DS during a tea party activity. However, during a puzzle activity, both groups of mothers used similar proportions of suggestive requests. The authors propose that mothers of children with developmental disabilities may offer guidance during a task differently than mothers of TD children. Although the current study did not include a TD comparison group, it did examine differences between adolescents with FXS-O and FXS+ASD. Between groups, maternal requesting behavior did not differ. That

is, there was no association between ASD group and maternal use of direct versus indirect, transparent versus ambiguous, or conventional versus unconventional requests.

Context familiarly and action norms may affect comprehension of requests, as young children do well with routinized requests (i.e., *your turn, put away the toys*) and with action requests rather than informational requests (Ervin-Tripp & Gordon, 1986). Similarly, many individuals with higher autism symptomology prefer routine and may have difficulty with change in routines. As such, for these individuals, context and routine familiarity may help clarify task expectations and provide desired structure to an activity which could in turn lead to increased comfort with the task and increased compliance. If the activity is one in which the routine is deeply engrained or one with which the adolescent is familiar, then maternal requesting behavior may not influence compliance, as the context necessitates certain actions more so than the mother's requesting.

The current study examined adolescent compliance during a 10-minute mother-son snack making task. The dyad was provided with materials and step-by-step instructions on how to make the snack together. As such, this interactive context was fairly structured which may have influenced the mothers' directiveness during the activity (Landry et al., 1994). In a study of young children with DS, a more directive requesting style, in contrast to a suggestive requesting style, was associated with increased compliance during an unstructured task (tea party) but not a structured one (puzzle) (Landry et al., 1994). Thus, the authors suggest, children may benefit more from directive guidance during unstructured tasks and from suggestive guidance during structured ones, as the social expectations are not as clear in unstructured tasks. In the current study, the snack task was highly structured to the extent that mothers may have believed there was a single correct way to make the snack, which may have led the mothers to believe that the

task had to be completed exactly as the instructions listed. However, as in the study by Landry and colleagues (1994), mothers may have relied on the inherent task structure to guide their sons' actions. On average, mothers used more direct requests than indirect ones, suggesting they used directive guidance to help their sons through the activity rather than suggestive guidance. Future studies would benefit from the addition of another interactive context that would be less structured and more conducive to following the son's lead, such as doing a puzzle. It is likely that maternal requesting interacts with contextual demands to impact compliance.

Adolescent Characteristics

In the FXS group, compliance was not associated with request directness, transparency, or conventionality, but it was associated with adolescent characteristics such as language, social skills, and autism symptomology. Individual differences in language ability, as captured by measures of receptive vocabulary, expressive vocabulary, talkativeness, and nonverbal cognition, were predictive of likelihood of compliance following a maternal request for action. These findings contrast with those of Kissine and colleagues (2015; 2012) and Bryce and Jahromi (2013) who did not find associations between compliance and nonverbal cognition and receptive language in ASD, respectively. This study considered receptive vocabulary not receptive syntax as a predictor of compliance. In a study of TD 4-year-olds, Ledbetter and Dent (1988) demonstrated that children with higher receptive syntax abilities were more compliant with complex requests (i.e., those with two verbs, such as *Can you put the spoon where the pudding is?*). An assessment of receptive morphosyntax would strengthen this study by determining the types and complexity of structures that the participants understood.

In addition to considering receptive syntax abilities, an examination of expressive syntax would be informative. Primarily, characterizing the expressive syntactic abilities of adolescent

males with FXS and ASD would show whether or not they produce the types of requests considered in this study. If they use requests for action directed towards their parents, siblings, or peers then it would be expected that they can comprehend these requests as well, given that comprehension precedes production. An analysis of productive request structure and relative frequency would also inform how adolescent males with FXS or ASD perform compared to developmental expectations for requesting. Ervin-Tripp and Gordon (1986) suggest that between four and eight years-old, children become effective requesters and are able to successfully gain attention, maintain appropriate social relationships, and ensure clarity and persuasion in their requests. Males with FXS have known delays in expressive syntax (Price et al., 2008; Sterling, 2009, 2018), but little is known about their requesting behaviors, especially as pertains to directness, transparency, and conventionality.

Autism Symptoms

This study builds on previous research by considering the impact of autism symptoms (both as a categorical diagnosis and as a continuous metric) on compliance. When differentiating the adolescents with FXS by autism symptomology, those with higher autism symptoms were significantly less compliant on average. Bryce and Jahromi (2013) also found differences in compliance by autism status in their study of TD and ASD children. In the current study, participants with FXS and higher autism symptom severity tended to have lower language ability, lower social skills, and lower nonverbal cognition. All three of these were associated with compliance, so it may be that there is a complex mediating relationship between autism symptoms, linguistic and social ability, and compliance in adolescent males with FXS. Indeed, elevated autism symptom severity or co-morbid diagnoses have been repeatedly associated with lower performance in social communication and language in this population (e.g., Flenthrope &

Brady, 2010; Klusek et al., 2014; Lorang & Sterling, 2017; Losh et al., 2012; Martin et al., 2017; Sterling, 2018).

When examining the differences between adolescent males with FXS+ASD and those with FXS-O, two methods of measuring autism symptom severity were used. The adolescents were first grouped into diagnostic categories based on their CARS2-ST scores and then they were also evaluated based on their total CARS2-ST scores, which was a continuous variable rather than a categorical diagnosis. For both methods of measurement, adolescents with more severe autism symptoms were less likely to comply. However, the categorical predictor was stronger than the continuous one; the categorical predictor had a higher effect size and accounted for a higher proportion of the variance in between-person differences. This was unexpected because it was initially hypothesized that the continuous predictor would capture individual differences in autism symptomology between persons more accurately than the categorical one. Abbeduto et al. (2014) suggested that continuous measures of autism symptom severity in FXS may be preferred over categorical ones because categorical diagnoses can lead to clinically relevant and nuanced symptoms being overlooked. This recommendation may still be true, as the differences between the two measurement methods in this study were small. Furthermore, this study relied on a single assessment of autism symptoms, rather than combining assessments to arrive at a diagnosis or considering multiple assessments of autism symptom severity. For example, use of the Autism Diagnostic Observation Schedule (Lord et al., 2012) may have yielded different groups than the CARS2-ST, an issue that has recently come into consideration (Abbeduto et al., 2019; Fielding-Gebhardt et al., 2021; Haebig et al., 2020).

Request Comprehension

Request comprehension was measured by compliance with the request. Thus, this study operated under the assumption that comprehension of the request is followed by compliance and that if the adolescent did not comply, then he did not comprehend the request. Given that this was a study of adolescents with known cognitive delays and high rates of maladaptive behaviors, it cannot be concluded that request comprehension is the same as compliance. Several methods could better characterize language comprehension in this population. Comprehension could be considered following multiple of the same or similar requests under the assumption that repeated requesting is necessitated by poor comprehension. Additionally, comprehension could be considered as a function of latency to comply following the request with the assumption that a longer time between the request and the compliant response may indicate processing speed. However, these two methods still assume that the adolescent would comply if they comprehended the request. A stronger method of characterizing language comprehension would be by studying eye-gaze. First, eye-gaze could help determine comprehension by tracking the participant's gaze during and after a request. Regardless of compliance, an eye-gaze paradigm could determine whether or not the participant comprehended the request, as the timing of their looking and orienting to an object in the request would inform their understanding of that request. Studies of this nature have been useful in examining comprehension of garden-path sentences in typically developing children and adults (Tanenhaus et al., 1995; Trueswell et al., 1999).

A lag sequential analysis, or one in which a sequence of behaviors is examined, may be another informative way to consider compliance with requests. This type of analysis would include antecedents and consequences of behaviors. For example, a mother may request the

adolescent perform an action, then the adolescent may be noncompliant, and then the mother may request that action again at which point the adolescent is compliant. In this study, maternal requests were analyzed on a turn-level rather than an utterance-level, meaning that the mother could have spoken multiple utterances to request a behavior and that compliance was associated with the last utterance of the turn, not each individual utterance. A fine-grained analysis where each utterance is considered as an antecedent, rather than each turn (so "put away the trash" versus "put away the trash. Go ahead. Put it over there.") could have different outcomes, as adolescent responsivity to utterances may differ from their responsivity to turns. Additionally, future studies could use this methodology to examine incremental comprehension with multi-step or multi-clause requests. For instance, if a child were told "*first put the trash in the bin and then take the bin outside*", fine-grained consideration of their behavior may be more indicative of their comprehension than the turn-level coding that was used for this study.

ASD Group

The inclusion of an ASD group was initially proposed for this project so that adolescents with FXS-O, FXS+ASD, and idiopathic ASD could be compared to one another on compliance as well as language and social skills. This three-group comparison is common in the field (Klusek et al., 2014; Lee et al., 2016; Smith et al., 2012), and is a means to better understand the FXS phenotype and how it relates to ASD. Several studies have compared these groups on language and social communication skills (Klusek et al., 2014; Lee et al., 2016) or on behavioral phenotypes (Smith et al., 2012). Other studies have compared language in individuals with idiopathic ASD or FXS-ASD and found that the two groups differed in profiles of language ability (Haebig & Sterling, 2017), with boys with FXS-ASD demonstrating impaired grammar (Sterling, 2018). In general, it has been demonstrated that impairments in social communication

are a shared feature between FXS-ASD and idiopathic ASD (Lee et al., 2016), and that individuals with FXS-ASD may have significantly different performance on measures of pragmatic language (Martin et al., 2017) than the other two groups.

Unfortunately, this study was underpowered to make such comparisons due to difficulty recruiting participants and collecting data during the COVID-19 pandemic. The two participants in the ASD group demonstrated different profiles of compliance and language and social skills, as would be expected given the heterogeneity of ASD. Both adolescents had limited verbal communication, and one performed poorly on the PPVT-4 due to noncompliant behaviors while the other completed the assessment. Both demonstrated deficits in social skills, as expected given their diagnoses. When comparing the adolescents with ASD to those with FXS (-O or +ASD), performance across measures was similar. With the exception of one participant's PPVT-4 raw score, both adolescents in the ASD group had scores on the assessments and language sample that were within the range of the FXS group. Expansion of the ASD group would be beneficial so that meaningful comparisons and conclusions can be drawn.

Limitations

As discussed, there are several ways in which this study could be expanded upon in the future. Similarly, there are several limitations. First, the method used to measure compliance relied on video recordings of the mother and adolescent. There were times when the adolescent would leave the task area, and so compliance would either be scored as *no opportunity* (and thus removed from the analysis set) or compliance would be scored based on benefit of the doubt and the mother's reaction to the adolescent. It would be beneficial to include an additional camera angle to capture times when the adolescent left the task area, but this was impractical given the resources available for this study. Additionally, perseverative language use is high in individuals

with FXS (Murphy & Abbeduto, 2007), and the utterances used to measure MLUm and number of utterances included instances of perseveration. This limits characterization of the adolescent's generative syntax and language, as repetitive phrases may not adequately capture their morphosyntax or talkativeness. However, including the perseverations is an accurate representation of daily language use in this population. As is often the case with studies of neurodevelopmental disorders, the sample size of this study was small. A larger sample would have allowed for more sophisticated quantitative modeling, specifically the inclusion of multiple predictors. Furthermore, a larger ASD group would allow for more sophisticated comparisons between groups. Finally, while previous work in this area has largely focused on toddlers and young children (Landry et al., 1994; Ledbetter & Dent, 1988; Rimac, 1985; Rocissano et al., 1987), this study focused on adolescents. Because a TD group was not included as a control (either MA or CA matched) it would be difficult to infer whether noncompliance is due to failure to comprehend or whether it is due to attitudes and behaviors that are part of adolescence.

Implications

Maternal language use may influence how children complete actions. As demonstrated by Rocissano et al. (1987), TD toddlers are more likely to comply when the request follows the child's lead and is on topic. Similarly, Landry et al. (1994) found that young children with DS have higher rates of compliance when the maternal request follows their lead. Additionally, Lemanek et al. (1993) found that preschoolers with ASD, intellectual disability, language impairment or TD were more compliant when requests were positively worded and clear, rather than when they were negatively worded or gestural. Finally, Landry et al. (1994) concluded that mothers can aid in child compliance when they clarify the structure and expectations of a task,

particularly one that is novel or unstructured. As such, it may be beneficial to child compliance if the mother requests in certain ways.

It was once believed that children with language delays should receive simpler language input than their TD peers. Specifically, Blue (1981) argued that individuals working with children with language delay should avoid using indirect requests and ambiguous statements. However, the results of this study suggest otherwise. Indirect, ambiguous, or unconventional requests were not seemingly harder to comply with than their counterparts. Focusing on indirect requests, this style of requesting is suggestive, may provide an illusion of choice to the adolescent, and is a gentler, and often more polite, way of requesting. This type of request may promote autonomy by providing choice and structure without commanding or ordering the adolescent to do an action. Although it has been hypothesized that children with language delays may benefit from avoiding indirect requests, this is not borne out in this or other studies (Kissine et al., 2015; Kissine et al., 2012; Rimac, 1985).

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Appendix A: Scoring Manual for Maternal Request Properties

All utterances are segmented by C-units and multiple requests for action that immediately follow one another are coded on the same line, therefore, it is possible to have several requests occur during the same time-stamped event. Only the final request is coded for child compliance. Consequently, only the final utterance will be coded as direct or indirect. See exceptions under Indirect Request for Action # 7 below.

Directiveness

Refers to the degree to which the intended meaning of the request for action is explicitly stated.

Direct Requests for Action

Direct requests are "explicit statements specifying an action" (Bryce & Jahromi, 2013; pp. 239). A direct request is always transparent and conventional, see below.

1. Imperatives (i.e., commands) that are complete sentences, for example:
    a. M Come here.
    b. M Sit down.
    c. M Stop.
    d. M Wait.
    e. M Look.
    f. M Put the pudding in the cup.
    g. M Don't do that.

Note. Imperatives may contain a second person subject (i.e., you or the child's name), for example:

    h. M You go first.
    i. M Sarah, come back.

2. Imperatives with an elided "Be" verb which, therefore, consist of only an adjective. The following list is exhaustive:
    a. M Careful.
    b. M Quiet.
    c. M Gentle.
    d. M Nice.

3. Statements with a second person subject (i.e., You) and one of the following verbs paired with an infinitive: *need, must, have, have got, got*. For example:
    a. M You need to sit down
    b. M You must slow down.
    c. M You have to come here.
    d. M You gotta do this first.
    e. M You've got to shake it.

4. Sentence fragments that **immediately** follow a direct request (i.e., occur on the same line):
    a. M Come here. M right here.

b. M Stop. M No, John.
c. M Give it to me. M hey, now.

Note. The following example should be coded as **indirect** because the sentence fragment (i.e., M right now) does not immediately follow the direct request (i.e., M give it to me); there is an intervening indirect request (i.e., M I need that one).

d. M Give it to me. M I need that one. M right now.

Indirect Request for Action

Indirect requests are polite requests that offer suggestions, choices, or guidance, rather than explicitly specifying an action (Bryce & Jahromi, 2013). An indirect request may be transparent or ambiguous and conventional or unconventional.

1. Abandoned utterances that are incomplete, including abandoned imperatives, for example:
    a. M Put that>
    b. M Take the>
    c. M You need to>

2. Statements that are merely suggestive or indicate that the child has a choice, including imperatives paired with "maybe". Generally, these will have been accompanied by a gesture.
    a. M Maybe put them in your hands and crush them.
    b. M You can sit right here.
    c. M It's your turn.
    d. M I need your help.
    e. M I want you to pour it into the big bottle.
    f. M You might want to crush the cookies in your hands.
    g. M You're supposed to crush the cookies with your hands.

3. Utterances that contain "Let's…" or "We…"
    a. M Let's read the directions first.
    b. M Let's try again.
    c. M Maybe we should clean up first.
    d. M We need to crush the cookies now.

4. Implicit requests for the child to read, for example:
    a. M What's the next step?

5. Requests that consist of only a gesture (Note: there will be no transcription, just a place holder to indicate the mother's turn in the conversation).
    a. M .
    b. M *points*.

6. Questions (except those that pertain to reading).
    a. M Can you sit down?
    b. M Will you get the milk?

c. M Push that chair over, will you?
d. M Where should that go + *gesture*?
e. M What do you do with the milk + *gesture*?

7. Utterances that do not contain a verb (i.e., sentence fragments) other than those utterances with an elided BE verb listed under #2 of Direct Request Behavioral Comply **AND** do not occur with an **immediately preceding direct request behavioral comply** on the same transcription line, for example:

a. M Hey Hey.
b. M right here.
c. M No.
d. M Your turn.

These utterances will have been accompanied by a gesture that supplanted the verb.
Note: Sentence fragments that **immediately** follow a direct request (i.e., occur on the same line) should be coded as **Direct Request Behavioral Comply**:

e. M Come here. M right here.
f. M Stop. M No, John.
g. M Give it to me. M hey, now.

Note: The following example should be coded as **indirect** because the sentence fragment (i.e., M right now) does not immediately follow the direct request (i.e., M give it to me); there is an intervening indirect request (i.e., M I need that one).

h. M Give it to me. M I need that one. M right now.

Transparency

Transparency refers to the surface structure specification of the agent, action, and object (optional).

Syntactic Structure and Semantic Roles: Every sentence, minimally, is made of an action and a doer of that action. The action is the verb, and the agent is the entity who performs the action. Depending on the verb, there may or may not be a direct object (and an indirect object).

Transparent Request for Action (score = 1)

A transparent request for action is one in which the agent (intended doer of the action) and the action (a verb that can be performed) are overtly specified in the syntactic structure. It is important to note, in English, the agent of an imperative statement (you) is not always explicitly stated. Rather, in imperative statements, English allows for the agent to be elided.

If the action is transitive (e.g. Eat the apple) or ditransitive (e.g., Put the apple on the table), the request is only transparent if the object(s) follows the action. Objects can be substituted with pronouns and determiners (it, that, one, etc.) which take the place of the noun. Locatives can be shortened too (e.g. Put it in here vs. Put the apple in the trash can).

Examples include:

a. M Johnny, pick a game.
b. M put away the trash.
c. M I need you to close the door.

d. M do you mind moving the puzzle piece?
e. M Please flush the toilet, James.
f. M keep going.
g. M you_do_it [sign]. **signs are considered transparent for children who use sign as their primary means of expressive communication.*
h. M pick it up.
i. M grab one from there.
j. M give me that.
k. M stir it.

<u>Ambiguous Request for Action (score = 0)</u>

An ambiguous request for action is one in which the agent and action are not overtly specified. The agent may be unclear, inanimate, or ambiguous or they may not specify an action or an object. Gestures are considered ambiguous requests.

Unclear

a. M someone needs to clean the counter.
In this case, the agent is not specified as a single person. The agent is a broad term.

Inanimate

a. M the counter needs to be cleaned.
Here, an inanimate object requires an action to be performed on it, but the agent is not specified.
b. M that block goes on top.
Here, the request does not specify an agent.

Ambiguous: One or more persons in the interaction can complete the action.

a. M let's clean the counter.
In this case, the mother and/or the child could be the agent.
b. M we'll shake.
Unclear whether mother or child is the expected agent.

No Action specified

a. M your turn.
b. M like you mean it.
** the action is not specified here.*
c. M you next.
d. M come on.
** the action is not specified here.*
e. M what does the next one say?
** There is no specific agent and action- requires child to infer mother's intent*

Transitive action lacking object(s)

a. M put some of this^
b. M can you cover^
c. M give me the^
d. M make them^

Gestures

a. In this coding system, all gestures are considered ambiguous because gesture morphology was not specified during transcription for the FXS group.

Conventionality

Conventionality refers to the degree to which a request for action can be recognized within the social communication expectations of a culture.

Conventional Request for Action (score = 1)

A conventional request for action is always interpreted as a request. The request can be interpreted as such in the absence of contextual cues. A conventional request is clearly a command.

a. M come inside.
b. M I need you to put the box away.
c. M can you bring me the milk?
d. M keep going.
e. M let's turn on the iPad.
f. M your turn. **this is conventional because it is within social expectations for signaling an action during a game or sequence.*
g. M all_done [sign]. **signs are considered conventional for children who use sign as their primary means of expressive communication.*

Unconventional Request for Action (score = 0)

An unconventional request for action can be interpreted as a suggestion, statement, preference, or opinion. The request is unlikely to be interpreted as a command without informative contextual cues. An unconventional request is a passive suggestion.

a. M the door needs to be closed.
b. M that block goes on top.
c. M if you don't mind right now, can you grab my purse?
d. M are you able to pass the salt?
e. M is it possible for you to cover your face?
f. M while I mix the pudding, how about you read the next one?
g. M like you mean it. **this needs context to make sense.*
h. M come on. * *this requires contextual information to make sense*
i. M why don't you take a try?

In this coding scheme, t**ransitive actions lacking object(s)** are considered unconventional because contextual cues would be needed to deduce the entire meaning of the request.

j. M put some of this^
k. M can you cover^
l. M give me the^
m. M make them^

In this coding scheme, all **gestures** are considered unconventional because gesture morphology was not specified during transcription for the FXS group.

Appendix B: Scoring Manual for Requests for Action and Child Compliance

Maternal Requests for Action

A non-verbal or verbal request for behavioral comply is a directive to which the child can comply behaviorally. It must fall into one of the following categories:

<u>An imperative statement (must contain a verb).</u>

a. M you do it.
b. M get the milk.
c. M sit down. M and read the book.
d. M don't do that.
e. Abandoned utterances that are imperatives are coded as request for behavioral comply not comment.
    i. Now find the red that just has>

Note: This does not include imperative-like statements that function as comments. (For example 24041-7_pzl 199sec: M oh look. M this one's up your alley. M look. M you can poke it out the side).

"yes" and "no" responses that are immediately followed by an additional utterance are transcribed as separate c-units but kept on the same line as the rb modifier. (e.g., M no. M stop that.). Do not place yes/no responses on a different line with a comment modifier.

<u>An indirect request</u> with a clear intent for the child to behaviorally comply that may include the following verbs (will, can, need, should, could, let's, might, why don't, how about, got to [meaning "have got to"]). ***Focus on the mother's expectation not just the words she uses.***

a. M you need to sit down.
b. M you should sit down.
c. M why don't you get that out.
d. M why don't you pull that one.
e. M let's start shaking. \hands the bottle to the child and looks at him expectantly\
f. The orange one is next \hands the child a puzzle piece\.
    a. Code as a single event (RBC), not as a comment followed by a RBC. For example: M The orange one is next \+ gesture\.
g. M I need your help \expectant look\,
h. M can you show me?
i. M will you do it?
j. M Let's do it. Let's try. Let's go. Let's put this on. Let's read this.
k. M Let's see if we can do this one.
l. M let's not put it there.
m. M let's not open it.
n. M can you twist it on good?
o. M other way!

p. M uhuh! (to indicate 'stop what you are doing')

The following are examples of comments, rather than indirect requests for behavioral comply:

a. Let's see. \as a stand alone sentence\
b. We're not gonna put it away (without accompanying gesture).
c. We're gonna make this castle.
d. You're not doing that one yet.
e. You're not getting my phone.
f. Hold on. Let me see (with no clear expectation for the child to stop or start an action).

Note: If you are unsure if the mother's utterance is a request for behavioral comply or a request for verbal comply default to request for verbal comply.

If you are unsure if the mother's utterance is a comment or an indirect request for behavioral comply, default to comment unless there is an accompanying gesture from the mother which indicates "no, stop, wait" (such as removal of item or restriction of behavior).

If the mother is narrating her own actions, this gets coded as a comment, not a request for behavioral comply because there is no expectation that the child perform the action. Most often, this occurs with a "let's" statement from the mother. E.g., "M let's take the pieces apart".

A gesture to which the child can behaviorally comply such as give, distal/proximal point, tapping, and show. Again, the gesture must be paired with an expectant pause. This may appear as a corrective demand, such as in 02081-7_pzl time 337.60 where in the mother points to the puzzle book and clearly expects the child to correct his puzzle. If the mother has a request behavioral comply that is purely gestural (no co-occurring verbalization) transcribe M . to mark her turn in the conversation.

a. Mother requests that the child sit down by looking at the child and pointing to the chair.
b. Mother requests the child shake the bottle by looking at the child and holding the bottle towards him/her.
c. Mother looks at the child and gives him trash to throw away.
d. If the mother requests that the child give her a high five and holds her hand up this is coded as a request behavioral comply only when it is not paired with a mother child-directed positivity (either through words indicating positive judgement or through positive tone).
    a. Code these as a request behavioral comply:
        i. M where's my high five \+ holds hand for high five\?
        ii. M give me a high five \not paired with a positivity\.

b. Code this as a mother child-directed positivity:
    i. M good job. M give me five.
    ii. M awesome! M high five!
    iii. M give me five \+ overt positive tone\.

Note: If the child high fives the mom in response to her request (behavioral request or just holing her hand up along with a positive statement) this is always coded as a gesture.

Hand-over-hand manipulation. When the mother uses hand-over-hand guidance to complete an action this is coded as a request for behavioral comply.

a. Mother takes the child's hand to activate the iPad. If the mother continues to grasp the child's hand for an extended period of time and is manipulating the hand to complete a specific task, this only counts as one request behavioral comply. If the mother makes an intervening comment, child-directed positive statement, or request verbal comply **while** manipulating the child's hand, the utterances are transcribed and coded appropriately on separate lines. If the child pulls away and the mother takes the child's again, this counts as another request behavioral comply.

Direct requests or imperatives for the child to read, either to themselves or aloud, default to requests for behavioral comply.

i. Read this.
ii. I need you to read this.
iii. Can you try reading this?
iv. Can you read it to me?
v. What does this say? (as the Mom is indicating something on the recipe card or iPad)
vi. Verbal prompts to complete a sentence as the Mom is indicating something on the recipe card or iPad (e.g., Pour~)

Note: "Your turn" is always considered a request behavioral comply.

If the tape ends after a request behavioral comply and the child does not have a chance to respond to that request (i.e., there is no opportunity to determine child's compliance) then default this code to comment.

Child Compliance

Please note, compliance codes must be time-stamped as occurring after the request for behavioral compliance has been spoken by the mother. Do not put a compliance code in the Noldus transcript before the mother has finished speaking the request for behavioral compliance. When there are multiple requests for action within 3 seconds of each other, only consider compliance to the last request for action.

Compliant

A child is compliant when the child responds in a socially appropriate manner to the mother's request for behavioral comply.

The request does not need to be fully understood by the child, as long as an appropriate effort to comply is made.

a. M Let's crush the cookies.
   Child proceeds to grab the bag of cookies and crush them with his/her hands.
b. M Hand me the iPad please.
   Child politely hands the mother the iPad.
c. M Can you put the green block in the middle?
   Child proceeds to put the green block in the middle right away.
d. M Please give me the carton of milk.
   Child looks for the milk but hands the mother the pudding mix.
e. M Now set the block right in front.
   Child sets the block the wrong way by accident.
f. M You pour the milk.
   Child pours the milk.

<u>Default to Compliant</u>

If it is unclear what the child's response should be, or if the mother's request falls into one or more of the following categories, code as compliant.

1. The mother's request is **irrelevant**. This may include giving a request that warrants no response from the child.
   a. M Wait wait wait wait.
      Child is already sitting patiently waiting.
2. The mother's request is an **abandoned utterance**.
   a. M Let's do>
   b. M Can we>
   c. M Put that one. M Try>
   d. M Now find the red block that just has>
   e. M Alright. M then we need>
3. The mother asked the child to do something he or she **already completed**.
   a. M Can you build the train tracks?
      C Mom, I already did that.
   b. M Put the puzzle piece right there so you can see>
      Child already placed the puzzle piece.
   c. M Turn it so the door faces right.
      C The door is already facing right.
   d. M Take all the little tops out of the castle.
      Child does not respond because he or she already did it.
4. The behavioral request is **incorrectly coded** by the coder. When this is the case, code as compliant.
   a. M wait wait. M where did I put the instructions?
      This does not warrant an action response from the child; it is a request for information.

Noncompliant

The child is noncompliant when:

1. S/he **ignores the mother's request or otherwise does not comply**.
   a. M Come here, please.
      Child continues to walk away.
   b. M hand me the gummy worms.
      Child ignores the mother and eats the cookies.
2. The mother's **request is interrupted** by the child.
   a. M So let's^
      Child vocalizes "No!" before mother is able to finish her request.
3. The child repeats the mother's request but **does not actually complete the request**.
   a. M Let's play this game. Push the button.
      C Verbalizes "push" but does not actually push the button.
4. The child appropriately or inappropriately **declines** the mother's request.
   a. M You do it.
      C can you do it?

No Opportunity

The child has no opportunity to comply when:

1. The mother gives the child a request yet immediately **completes it herself**.
   a. M Shake the bottle.
      Mother immediately shakes the bottle for the child.
   b. M Pull the puzzle apart.
      Mother immediately proceeds to pull the puzzle apart.
   c. M Ok well put the iPad down so we can>
      Mother grabs the iPad from the child before she finishes her request and/or before the child has a chance to put the iPad down.
2. The mother uses **hand-over-hand prompts** to assist the child and therefore does not allow the child the opportunity to independently complete the requested behavior.
3. The mother **does not give the child enough time** before the next utterance (or request) to appropriately respond. Code "no opportunity" for the first request behavioral comply if a comment occurs between two requests for behavioral comply.
   a. M Give me the pudding (behavioral request, 240.00)
      M You like chocolate pudding (comment, 241.5)
      [No opportunity].
      M Open it. (behavioral request, 243.00)
      C okay. (verbal, 245.00)
      [Child compliant].
      The first request would be coded as no opportunity and the second request would be either compliant or noncompliant.

General Guidelines for Coding Compliance

- If the coder cannot see the child's actions due to the setting or camera angle (example: pushing buttons on the iPad), a code is given based off of the mother's response.

    M Push the button.
    Child's action is not on camera/ is obscured by the iPad.
    M There you go. M good job.
        - This example would be coded under compliance due to the nature of the mother's response.
- Code the response at the **beginning** of the action.

    M Pour the milk
    Child begins to pour the milk→128.20
    Child finishes pouring the milk→132.4
        - Code the response at 128.20
- When there are multiple requests in a row that are all on the same event line (i.e., multiple requests for behavioral comply within three seconds of one another and with no other behaviors in between them) only **consider the child's response to the last request**.

    M You do it.
    M Make it like that.
    M Make it like that.
    M Wait wait wait.
    M Slow down.
    M Hang on.
    M Alright then hand me the timer.
        - Code the response to "alright then hand me the timer".
- If there are several requests for behavioral comply within a short time frame and the child compliance codes start overlapping, make sure to indicate in the comment section which event/request for behavioral comply the compliance code is in response to.

    M Hand me the orange puzzle piece. (behavioral request, 123.40)
    M Stop playing with your shirt. (behavioral request, 127.21)
    Child stops playing with his or her shirt. (compliant, 127.21)
    Child gives the mother the orange puzzle piece. (compliant, 123.40)