

**Temperament Predicts Challenging Behavior in Children with Autism Spectrum Disorder at
Age 5**

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Abstract

Background: Challenging behaviors during early childhood have a significant impact on cognitive and social development. The present study aimed to identify the developmental predictors of these behaviors in preschool aged children with Autism Spectrum Disorder (ASD) at 2-year follow-up. We examined temperament, which has been identified as key to emotion regulation in typical development, as well as developmental level and **ASD symptom severity**, as potential predictors of parent-reported challenging behavior.

Method: Forty-three parents of preschool aged children with ASD from a previous study were invited to participate. Data from 26 children with ASD aged 4 to 6 years ($M = 5$, $SD = .60$) were available for follow-up analyses. Developmental level, **ASD symptom severity**, and temperamental difficulty at baseline were considered as potential predictors of frequency and severity of challenging behavior at follow-up.

Results: Baseline negative affectivity was uniquely predictive of frequency of challenging **behavior at follow-up**. Although no individual variable was identified as a unique predictor of variance, the combined effects of temperament were predictive of the severity of challenging behavior at follow-up, contributing to 46% of variance in scores.

Conclusions: These findings highlight the potential impact of emotion-regulation related aspects of temperament on later emerging challenging behavior in young children with ASD, suggesting opportunities for early intervention. **Results also identified a role for developmental level in the severity of challenging behavior, but suggest that the effect may be metered by temperament.**

Keywords: autism; ASD; challenging behavior; developmental delay; temperament

Highlights

- Temperament, development and ASD symptom severity were examined as predictors of challenging behavior
- Negative affectivity predicted frequency of challenging behavior at follow-up
- No individual variables uniquely predicted severity of challenging behavior at follow-up
- Emotion regulation-related aspects of temperament may predict later challenging behavior in ASD

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Children with Autism Spectrum Disorder (ASD) are at greater risk of exhibiting challenging behaviors compared to their typically developing peers (Baker et al., 2003) and those with other developmental conditions (Matson & Nebel-Schwalm, 2007). Behaviors may include aggression to self or others, property destruction, elopement (Hattier, Matson, Belva, & Horovitz, 2011; Hill et al., 2014; Totsika, Hastings, Emerson, Lancaster, & Berridge, 2011), and can affect opportunities for socialization and learning (Matson & Nebel-Schwalm, 2007) and family quality of life (McStay, Trembath & Dissanayake, 2014). It is therefore important to identify risk-markers to inform interventions and to support the positive developmental trajectories of children with ASD. Although children more severely affected by ASD or who have significant cognitive or developmental delays are at greater risk of engaging in challenging behavior (Bronsard, Botbol, & Tordjman, 2010; Dominick, Davis, Lainhart, Tager-Flusberg, & Folstein, 2007; Jang, Dixon, Tarbox, & Granpeesheh, 2011; Matson, Wilkins, & Macken, 2008; Murphy et al., 2005), other early risk-markers have received less attention in the literature. One potential candidate is temperament difficulties, including children's difficulties regulating their self, including their emotions (Rothbart & Sheese, 2007; Rothbart, Ellis, & Posner, 2004). The present study examined ASD symptom severity, child temperament, and developmental delay, as potential predictors of challenging behavior, at a 2-year follow-up of children (aged approximately 5 years at follow-up) who had previously participated in a study examining emotion regulation (see Nuske et al., 2019; Nuske, Hedley, Tseng, Begeer, & Dissanayake, 2018; Nuske et al., 2017).

Temperament

Early temperament difficulties may increase engagement in challenging behavior in young children with ASD. Temperament refers to early emerging, biologically-based individual differences in attention, affect, and motor responses (Shiner et al., 2012). Three higher-order dimensions of temperament can be derived across prominent temperament models (Chetcuti, Uljarević, & Hudry, 2019; Mervielde & Asendorpf, 2000) that relate conceptually to the facets proposed to underlie personality in later-life (De Pauw & Mervielde, 2010; Shiner & Caspi, 2003). We refer to these using Rothbart and colleagues' terminology, given the theoretical alignment of this study's temperament measure. Negative affectivity, or emotional stability/neuroticism, is the propensity towards negative emotions including worry, frustration, sadness, and anger. Effortful control is a counterpart of conscientiousness and encompasses the ability to shift attention and adaptively inhibit one's emotions and dominant responses. Surgency-extraversion, which is considered a counterpart of extraversion in personality taxonomies, refers to sociability versus social inhibition and energy/activity level (Evans, & Rothbart, 2007; Rothbart & Bates, 2006). Negative affectivity and effortful control have been linked with a variety of problematic behaviors in general population samples (for reviews, see De Pauw & Mervielde, 2010; Muris, & Ollendick, 2005; Shiner & Caspi, 2003), including those subsumed under the broader externalizing domain (i.e., hyperactivity, aggression, and defiance; Eisenberg, Cumberland et al., 2001; Eisenberg, Sadovsky et al., 2005; Eisenberg, Valiente et al., 2009; Hartley, Sikora, & McCoy 2008; Mazefsky et al., 2013; Supplee, Skuban, Shaw, & Prout, 2009). Findings regarding the impact of surgency-extraversion on challenging child behaviour are more varied (for a review, see Davis & Suveg, 2014), such that lower (e.g., Kim, Walden, Harris, Karrass, & Catron, 2007) and higher (e.g., Degnan et al., 2011) levels have been related to child externalizing problems.

To date, research on temperament in the context of ASD has focused predominantly on the characterization of group-level differences in relation to comparison groups (Chetcuti et al., 2019). This evidence suggests that ASD may be associated with an overall profile of lower surgency-extraversion and effortful control, and higher negative affectivity (Lodi-Smith, Rodgers, Cunningham, Lopata, & Thomeer, 2019). Prospective studies of infant siblings of children with ASD have also reported this temperament profile among those who do (versus do not) go on to ASD diagnostic outcome (Garon Bryson et al., 2009; Garon, Zwaigenbaum et al., 2016). However, fewer studies have examined relations between temperament and challenging behavior in the context of ASD (Chetcuti et al., 2019). One study in 2 to 8-year-old children with ASD found significant correlations between challenging behavior, higher negative affectivity and lower effortful control and surgency-extraversion (Adamek et al., 2011). Similar results have also been obtained among school-aged children (De Pauw, Mervielde, Van Leeuwen, & De Clercq, 2011; Kamio, Takei, Stickley, Saito, & Nakagawa, 2018) and adolescents with ASD (Burrows, Usher, Schwartz, Mundy, & Henderson, 2016; Schwartz et al., 2009), although surgency-extraversion was not significantly correlated with challenging behavior in adolescence. However, in addition to the limitations associated with the cross-sectional

design of these studies, the impact of developmental delay on challenging behavior was not considered.

Developmental Level

The degree to which individuals translate broader developmental abilities (e.g., cognition, language and communication) into practical-skills may buffer against engagement in challenging behavior (Conroy, Dunlap, Clarke, & Alter, 2005). Numerous studies have examined relationships between challenging behavior and language and communication delay (Dominick et al., 2007; Lecavalier, Leone, & Wiltz, 2006; Murphy et al., 2005; Sigafoos, 2000), non-verbal IQ (Estes, Dawson, Sterling, & Munson, 2007), and adaptive functioning (Baghdadli, Pascal, Grisi & Aussilloux 2003). Findings generally suggest that more significant developmental delay is associated with increased presence of challenging behavior. However, most of these studies have relied on cross-sectional samples thereby limiting results (e.g., Baghdadli et al., 2003; Dominick et al., 2007; Lecavalier et al., 2006; Sigafoos, 2000; c.f. Estes et al., 2007; Murphy et al., 2005). Longitudinal or follow-up studies are therefore needed to identify predictive relationships between developmental delay and challenging behavior in children with ASD.

Current Study

The current study examined ASD symptom severity, temperament (negative affectivity, effortful control, surgency-extraversion) and developmental delay as predictors of parent-report challenging behavior in a 2-year follow-up study involving preschool aged children with ASD. We predicted that ASD symptom severity, temperamental difficulties and developmental delay, would predict greater frequency and severity of challenging behavior at follow-up.

Method

Participants

Forty-three parents of preschool aged children with ASD who had participated in a study on emotion regulation were invited to participate in the follow-up study two years later. Inclusion criteria included being diagnosed with ASD and meeting criteria on the Autism Diagnostic Observation Schedule, second edition (ADOS-2; Lord, Luyster, Gotham, & Guthrie, 2012). Parents of 26 children aged 2 to 4 years ($M = 3.30$, $SD = .68$) at T1 and 4 to 6 years ($M = 5.00$, $SD = .60$) at follow-up (T2) consented to participate in the study (see Table 1 for child demographics). In addition to ASD, four of the 26 children were reported to have developmental delay and one child was reported to have Fragile X. Behavioral and developmental data for these participants were reviewed but did not emerge as outliers; hence, data were retained for these participants.

[Insert Table 1 about here]

Materials

Developmental level was assessed with the *Mullen Scales of Early Learning, Early Learning Composite* (ELC; Mullen 1995). Diagnosis was confirmed with the *Autism Diagnostic Observation Scale, second edition* (ADOS-2; Lord et al., 2012); the standardized comparison score (CS; range 1–10; Gotham, Pickles, & Lord, 2009) was used to determine ASD symptom severity (higher scores indicate greater severity). The 'Early Childhood Behavior Questionnaire (ECBQ; Putnam, Gartstein, & Rothbart, 2006) factor scales, 'Negative Affectivity', 'Effortful Control', and 'Surgency-Extraversion', were used to assess temperament. Items are scored using a 7-point Likert-type scale with higher scores for negative affectivity and surgency-extraversion, and lower scores for effortful control reflecting greater difficulties, respectively. In the present study internal consistency was reasonable for Negative Affectivity ($\alpha = .83$) and Effortful Control (reverse scored with higher scores reflecting greater difficulty; $\alpha = .72$), but not for Surgency-Extraversion ($\alpha = .36$). The *Home Situations Questionnaire, Pervasive Developmental Delay* (HSQ-PDD; Chowdhury et al., 2010) was used to assess frequency (dichotomous yes/no response) and severity (9-point Likert-type scale: mild = 1, severe = 9) of challenging behavior ($\alpha = .95$).

Procedure

The study was approved by the University human ethics committee. At baseline (T1), research sessions took place at the University child development center. Informed consent was obtained from parents while the child engaged in 5–10 minutes of warm-up play. Following the warm-up, parents completed questionnaires while their child engaged in a variety of experimental tasks from the larger study. If ADOS-2 or recent Mullen scores were unavailable, these assessments were

administered at the end of the session; average time between Mullen administration and T1 was 2.81 months ($SD = 3.60$, Range = 0–11.97), and ADOS administration and T1 was 7.97 months ($SD = 8.17$, Range = 0–31.2). At follow-up (T2; approximately two years following T1), parents completed a second consent form, additional demographic information and questionnaires. All questionnaires were administered online via the Qualtrics survey platform (Qualtrics, 2017).

Data Preparation and Analysis

Missing data analysis revealed that 1.03% (Range 0–7.7%) of data were missing for any single item. Five participants (19.23%) were missing some data (four items on the HSQ-PDD were missing 3.8% of data and four items were missing 7.7% of data). Little's MCAR test indicated data were missing at random, $p = 1.00$. Given pairwise and listwise exclusions would further reduce power for analyses thereby increasing the risk of Type II errors (Newman, 2014), missing values were replaced using multiple imputations in SPSS. Subsequent analyses utilized pooled imputed data. Spearman's rank order correlations were first used to examine the relationships between variables, and were used to inform model entry into hierarchical regressions for further analysis (please refer to Supplementary Materials for additional information regarding assumption checks and additional analyses; <https://www.doi.org/10.26181/5d28087098c9b>).

Results

Demographics

Demographics are provided in Table 1. At baseline (T1), Mullen ELC scores show that, on average, children were developmentally delayed. ADOS-2 CS ranged from 2 to 10 suggesting significant variability in ASD symptom severity between children. At follow-up (T2) children were typically involved in multiple therapies ($M = 3.84$, $SD = 1.90$). The overall sample was characterized as moderately to frequently engaged in challenging behavior on the HSQ-PDD, with these behaviors being rated as occurring at low to moderate severity.

Predictors

Table 2 provides correlations between baseline child characteristics (i.e., developmental level, autistic traits, temperament), and the frequency and severity of challenging behavior. Predictors of frequency and severity of challenging behavior were examined separately.

[Insert Table 2 about here]

Frequency of Challenging Behavior. Frequency of challenging behavior assessed with the HSQ-PDD at follow-up was strongly positively associated with baseline Negative Affectivity ($p < .001$) and negatively with Effortful Control ($p = .024$), but not Surgency-Extraversion ($p = .056$). Additionally, frequency of challenging behavior was not found to be significantly associated with either baseline developmental level assessed with the Mullen ELC ($p = .838$), or with autistic traits assessed with the ADOS-2 ($p = .195$). Next, a linear multiple regression model was applied to examine the unique contributions of Negative Affectivity (T1) and Effortful Control (T1) to Frequency of Challenging Behavior (T2; see Table 3). To control for developmental level, Mullen ELC scores were entered at Step 1 (refer to Table 3) and temperament variables were entered at Step 2. Step 1 accounted for 0.5% of the variance, $F(1, 24) = .119$, $p = .733$. Step 2 accounted for an additional 35.8% of variance, with the full model accounting for 36.3% of variance, $F(3, 22) = 4.19$, $p = .017$; however, only Negative Affectivity significantly and independently predicted the frequency of challenging behavior.

[Insert Table 3 about here]

Severity of Challenging Behavior. Severity of challenging behavior assessed with the HSQ-PDD at follow-up was strongly positively associated with baseline Negative Affectivity ($p < .001$) and negatively with Effortful Control ($p = .001$), Surgency-Extraversion ($p = .001$) and Mullen ELC ($p = .033$). Severity of challenging behavior was not significantly associated with ASD symptom severity on the ADOS-2 ($p = .998$). A linear multiple regression model was applied to examine the unique contributions of Negative Affectivity (T1), Effortful Control (T1), and Surgency-Extraversion to Severity of Challenging Behavior (T2; see Table 4), controlling for developmental level. Mullen ELC scores were entered at Step 1 (refer to Table 4) and temperament variables were entered at Step 2. Step 1 accounted for 31.6% of the variance, $F(1, 24) = 2.61$, $p = .119$. Step 2 accounted for an additional 46% of variance, with the full model accounting for 56% of variance, $F(4, 21) = 6.68$, $p < .001$;

however, none of the temperament variables were found to independently predict the severity of challenging behavior.

[Insert Table 4 about here]

Discussion

The current study examined relationships between baseline ASD symptom severity, developmental delay, and temperament as potential predictors of the frequency and severity of challenging behavior in young children with ASD at 2-year follow-up. To our knowledge, this is the first study to examine the role of temperament in predicting challenging behavior in young children with ASD that is not reliant on cross-sectional methodology. Temperament difficulties associated with effortful control and negative affectivity were significantly correlated with both the frequency and severity of challenging behavior in children with ASD at 2-year follow-up. Additionally, developmental level and surgency-extraversion were found to be associated with severity of challenging behavior. Following further analysis, negative affectivity was identified as a unique predictor of the frequency of challenging behavior. No unique predictors of the severity of challenging behavior were identified, although effortful control approached significance ($p = .063$), and the combined effect of negative affectivity, effortful control, and surgency-extraversion added significant variance to the model predicting severity, over and above developmental level which was not identified as a significant predictor when examined independently.

Our findings tentatively indicate that challenging behavior in young children with ASD may be more likely to be associated with early temperamental difficulties relating to emotional reactivity/dysregulation than developmental level, or even ASD symptom severity. Given the role that emotion regulation plays in creating an “affective homeostasis” or a buffer from which children dynamically and flexibly manage their emotional experience, including everyday stressors (Chow, Ram, Boker, Fujita, & Clore, 2005; Diamond & Aspinwall, 2003), it is not surprising that difficulties in aspects of temperament relating to emotion regulation are predictive of challenging behavior. These findings are consistent with recent studies reporting an association of internal physiological stress with challenging behavior in children and adolescents with ASD (Goodwin, Mazefsky, Ioannidis, Erdogmus, & Siegel, 2019; Nuske et al., 2019), as well as cross-sectional studies comprising older school-aged children/adolescents with ASD that have linked externalizing problems with low effortful control and surgency, and high negative affectivity (Adamek et al., 2011; Burrows et al., 2016; De Pauw et al., 2011; Kamio et al., 2018; Schwartz et al., 2009). Overall, although based on a small number of participants, these results suggest that temperament may be an important characteristic to consider regarding the trajectory of challenging behavior in children with ASD. Importantly, while temperament has been often linked with cognitive, emotional and social development (Kagan & Snidman, 1991), this study fills a notable research gap regarding the role of temperament in the development of challenging behavior in this population.

Unlike Estes et al. (2007) and other cross-sectional studies (e.g. Dominick et al., 2007), our findings failed to identify a strong role for developmental level in the frequency and severity of later challenging behavior in children with ASD, particularly when temperament is considered. The finding that baseline ASD symptom severity was not associated with challenging behavior at follow-up is also inconsistent with some prior research (e.g., Dominick, et al., 2007), possibly reflecting methodological differences across the literature (e.g., our use of the ADOS-2 Comparison Score to assess ASD symptom severity).

Clinical Implications

Targeting temperament difficulties early on in young children with ASD may help to prevent the development of challenging behavior. Given that developmental delay was also associated with severity of challenging behavior (although notably not when including temperament in the model), it may be that if a child who is prone to negative affectivity who is able to harness developmentally appropriate skills and abilities (which broaden their repertoire of coping and problem-solving strategies), the behavior may de-escalate. Conversely, a child with higher negative affectivity who is not able to harness such development skills, including coping and problem solving, may have more severe behavioral outbursts (Kagan, 1997). Therefore, taking into account children’s developmental level, in addition to temperaments difficulties such as effortful control, negative affectivity, or surgency-extraversion, may help to inform intervention programming to prevent or reduce challenging behavior in children with ASD.

Limitations

The study was limited by the small sample size leading to increased risk of Type II error and sample-specific results. Future research with larger samples are required to confirm longitudinal predictions of temperament with challenging behavior in children with ASD. The use of the ECBQ also has its limitations as the factor structure of the instrument has yet to be assessed in children with ASD. While there has been some attention given to the psychometric structure of personality measures in the context ASD (Barger, Campbell, & Simmons, 2014, 2016), the temperament measures often utilized in ASD populations have not yet been subject to such examination (Chetcuti et al., 2019). This would be a useful direction for future work. We note, nonetheless, that the internal reliabilities of the scales reported here offer some support for their use in this population, possibly with the exception of surgency-extraversion due to low Cronbach's alpha. The use of parent report to assess temperament and challenging behavior was also a limitation as results may be affected by parent-report bias (De Los Reyes & Kazdin, 2005). The use of multi-measure methodology (i.e., behavioral, physiological, clinical assessment) is recommended to reduce shared method variance (Chetcuti et al., 2019). Finally, it will be beneficial in future to include a measure of baseline challenging behavior.

Conclusions

Understanding the developmental origins of challenging behavior in children with ASD is critical given the potential impact these behaviors have on the child's cognitive and social development, and on their family's quality of life. Our results identify a potential contribution of early temperamental difficulties on later emerging challenging behavior, particularly dimensions related to emotion regulation. Our findings also suggest a small role for developmental level in the severity of challenging behavior in children with ASD, but suggest this effect may be metered somewhat by temperament.

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Author Contribution

SK, DH, and HN designed the study. SK, DH, LC, ES, and HN collected the baseline data and SK collected the follow-up data. SK and DH analyzed the data. SK, DH, ES and HN drafted the original version of the article, and LC contributed to the revision. All authors critically reviewed the article and approved the final article as submitted.

Conflict of interest

SK declares that she has no conflict of interest. DH declares that he has no conflict of interest. LC declares that she has no conflict of interest. ES declares that she has no conflict of interest. HN declares that she has no conflict of interest.

Compliance with Ethical Standards

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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Temperament Predicts Challenging Behavior in Children with Autism Spectrum Disorder at Age 5

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Abstract

Background: Challenging behaviors during early childhood have a significant impact on cognitive and social development. The present study aimed to identify the developmental predictors of these behaviors in preschool aged children with Autism Spectrum Disorder (ASD) at 2-year follow-up. We examined temperament, which has been identified as key to emotion regulation in typical development, as well as developmental level and ASD symptom severity, as potential predictors of parent-reported challenging behavior.

Method: Forty-three parents of preschool aged children with ASD from a previous study were invited to participate. Data from 26 children with ASD aged 4 to 6 years ($M = 5$, $SD = .60$) were available for follow-up analyses. Developmental level, ASD symptom severity, and temperamental difficulty at baseline were considered as potential predictors of frequency and severity of challenging behavior at follow-up.

Results: Baseline negative affectivity was uniquely predictive of frequency of challenging behavior at follow-up. Although no individual variable was identified as a unique predictor of variance, the combined effects of temperament were predictive of the severity of challenging behavior at follow-up, contributing to 46% of variance in scores.

Conclusions: These findings highlight the potential impact of emotion-regulation related aspects of temperament on later emerging challenging behavior in young children with ASD, suggesting opportunities for early intervention. Results also identified a role for developmental level in the severity of challenging behavior, but suggest that the effect may be metered by temperament.

Keywords: autism; ASD; challenging behavior; developmental delay; temperament

Highlights

- Temperament, development and ASD symptom severity were examined as predictors of challenging behavior
- Negative affectivity predicted frequency of challenging behavior at follow-up
- No individual variables uniquely predicted severity of challenging behavior at follow-up
- Emotion regulation-related aspects of temperament may predict later challenging behavior in ASD

Temperament Predicts Challenging Behavior in Children with Autism Spectrum Disorder at Age 5

Children with Autism Spectrum Disorder (ASD) are at greater risk of exhibiting challenging behaviors compared to their typically developing peers (Baker et al., 2003) and those with other developmental conditions (Matson & Nebel-Schwalm, 2007). Behaviors may include aggression to self or others, property destruction, elopement (Hattier, Matson, Belva, & Horovitz, 2011; Hill et al., 2014; Totsika, Hastings, Emerson, Lancaster, & Berridge, 2011), and can affect opportunities for socialization and learning (Matson & Nebel-Schwalm, 2007) and family quality of life (McStay, Trembath & Dissanayake, 2014). It is therefore important to identify risk-markers to inform interventions and to support the positive developmental trajectories of children with ASD. Although children more severely affected by ASD or who have significant cognitive or developmental delays are at greater risk of engaging in challenging behavior (Bronsard, Botbol, & Tordjman, 2010; Dominick, Davis, Lainhart, Tager-Flusberg, & Folstein, 2007; Jang, Dixon, Tarbox, & Granpeesheh, 2011; Matson, Wilkins, & Macken, 2008; Murphy et al., 2005), other early risk-markers have received less attention in the literature. One potential candidate is temperament difficulties, including children's difficulties regulating their self, including their emotions (Rothbart & Sheese, 2007; Rothbart, Ellis, & Posner, 2004). The present study examined ASD symptom severity, child temperament, and developmental delay, as potential predictors of challenging behavior, at a 2-year follow-up of children (aged approximately 5 years at follow-up) who had previously participated in a study examining emotion regulation (see Nuske et al., 2019; Nuske, Hedley, Tseng, Begeer, & Dissanayake, 2018; Nuske et al., 2017).

Temperament

Early temperament difficulties may increase engagement in challenging behavior in young children with ASD. Temperament refers to early emerging, biologically-based individual differences in attention, affect, and motor responses (Shiner et al., 2012). Three higher-order dimensions of temperament can be derived across prominent temperament models (Chetcuti, Uljarević, & Hudry, 2019; Mervielde & Asendorpf, 2000) that relate conceptually to the facets proposed to underlie personality in later-life (De Pauw & Mervielde, 2010; Shiner & Caspi, 2003). We refer to these using Rothbart and colleagues' terminology, given the theoretical alignment of this study's temperament measure. Negative affectivity, or emotional stability/neuroticism, is the propensity towards negative emotions including worry, frustration, sadness, and anger. Effortful control is a counterpart of conscientiousness and encompasses the ability to shift attention and adaptively inhibit one's emotions and dominant responses. Surgency-extraversion, which is considered a counterpart of extraversion in personality taxonomies, refers to sociability versus social inhibition and energy/activity level (Evans, & Rothbart, 2007; Rothbart & Bates, 2006). Negative affectivity and effortful control have been linked with a variety of problematic behaviors in general population samples (for reviews, see De Pauw & Mervielde, 2010; Muris, & Ollendick, 2005; Shiner & Caspi, 2003), including those subsumed under the broader externalizing domain (i.e., hyperactivity, aggression, and defiance; Eisenberg, Cumberland et al., 2001; Eisenberg, Sadovsky et al., 2005; Eisenberg, Valiente et al., 2009; Hartley, Sikora, & McCoy 2008; Mazefsky et al., 2013; Supplee, Skuban, Shaw, & Prout, 2009). Findings regarding the impact of surgency-extraversion on challenging child behaviour are more varied (for a review, see Davis & Suveg, 2014), such that lower (e.g., Kim, Walden, Harris, Karrass, & Catron, 2007) and higher (e.g., Degnan et al., 2011) levels have been related to child externalizing problems.

To date, research on temperament in the context of ASD has focused predominantly on the characterization of group-level differences in relation to comparison groups (Chetcuti et al., 2019). This evidence suggests that ASD may be associated with an overall profile of lower surgency-extraversion and effortful control, and higher negative affectivity (Lodi-Smith, Rodgers, Cunningham, Lopata, & Thomeer, 2019). Prospective studies of infant siblings of children with ASD have also reported this temperament profile among those who do (versus do not) go on to ASD diagnostic outcome (Garon Bryson et al., 2009; Garon, Zwaigenbaum et al., 2016). However, fewer studies have examined relations between temperament and challenging behavior in the context of ASD (Chetcuti et al., 2019). One study in 2 to 8-year-old children with ASD found significant correlations between challenging behavior, higher negative affectivity and lower effortful control and surgency-extraversion (Adamek et al., 2011). Similar results have also been obtained among school-aged children (De Pauw, Mervielde, Van Leeuwen, & De Clercq, 2011; Kamio, Takei, Stickley, Saito, & Nakagawa, 2018) and adolescents with ASD (Burrows, Usher, Schwartz, Mundy, & Henderson, 2016; Schwartz et al., 2009), although surgency-extraversion was not significantly correlated with challenging behavior in adolescence. However, in addition to the limitations associated with the cross-sectional

design of these studies, the impact of developmental delay on challenging behavior was not considered.

Developmental Level

The degree to which individuals translate broader developmental abilities (e.g., cognition, language and communication) into practical-skills may buffer against engagement in challenging behavior (Conroy, Dunlap, Clarke, & Alter, 2005). Numerous studies have examined relationships between challenging behavior and language and communication delay (Dominick et al., 2007; Lecavalier, Leone, & Wiltz, 2006; Murphy et al., 2005; Sigafoos, 2000), non-verbal IQ (Estes, Dawson, Sterling, & Munson, 2007), and adaptive functioning (Baghdadli, Pascal, Grisi & Aussilloux 2003). Findings generally suggest that more significant developmental delay is associated with increased presence of challenging behavior. However, most of these studies have relied on cross-sectional samples thereby limiting results (e.g., Baghdadli et al., 2003; Dominick et al., 2007; Lecavalier et al., 2006; Sigafoos, 2000; c.f. Estes et al., 2007; Murphy et al., 2005). Longitudinal or follow-up studies are therefore needed to identify *predictive* relationships between developmental delay and challenging behavior in children with ASD.

Current Study

The current study examined ASD symptom severity, temperament (negative affectivity, effortful control, surgency-extraversion) and developmental delay as predictors of parent-report challenging behavior in a 2-year follow-up study involving preschool aged children with ASD. We predicted that ASD symptom severity, temperamental difficulties and developmental delay, would predict greater frequency and severity of challenging behavior at follow-up.

Method

Participants

Forty-three parents of preschool aged children with ASD who had participated in a study on emotion regulation were invited to participate in the follow-up study two years later. Inclusion criteria included being diagnosed with ASD and meeting criteria on the Autism Diagnostic Observation Schedule, second edition (ADOS-2; Lord, Luyster, Gotham, & Guthrie, 2012). Parents of 26 children aged 2 to 4 years ($M = 3.30$, $SD = .68$) at T1 and 4 to 6 years ($M = 5.00$, $SD = .60$) at follow-up (T2) consented to participate in the study (see Table 1 for child demographics). In addition to ASD, four of the 26 children were reported to have developmental delay and one child was reported to have Fragile X. Behavioral and developmental data for these participants were reviewed but did not emerge as outliers; hence, data were retained for these participants.

[Insert Table 1 about here]

Materials

Developmental level was assessed with the *Mullen Scales of Early Learning, Early Learning Composite* (ELC; Mullen 1995). Diagnosis was confirmed with the *Autism Diagnostic Observation Scale, second edition* (ADOS-2; Lord et al., 2012); the standardized comparison score (CS; range 1–10; Gotham, Pickles, & Lord, 2009) was used to determine ASD symptom severity (higher scores indicate greater severity). The ‘*Early Childhood Behavior Questionnaire* (ECBQ; Putnam, Gartstein, & Rothbart, 2006) factor scales, ‘Negative Affectivity’, ‘Effortful Control’, and ‘Surgency-Extraversion’, were used to assess temperament. Items are scored using a 7-point Likert-type scale with higher scores for negative affectivity and surgency-extraversion, and lower scores for effortful control reflecting greater difficulties, respectively. In the present study internal consistency was reasonable for Negative Affectivity ($\alpha = .83$) and Effortful Control (reverse scored with higher scores reflecting greater difficulty; $\alpha = .72$), but not for Surgency-Extraversion ($\alpha = .36$). The *Home Situations Questionnaire, Pervasive Developmental Delay* (HSQ-PDD; Chowdhury et al., 2010) was used to assess frequency (dichotomous yes/no response) and severity (9-point Likert-type scale: mild = 1, severe = 9) of challenging behavior ($\alpha = .95$).

Procedure

The study was approved by the University human ethics committee. At baseline (T1), research sessions took place at the University child development center. Informed consent was obtained from parents while the child engaged in 5–10 minutes of warm-up play. Following the warm-up, parents completed questionnaires while their child engaged in a variety of experimental tasks from the larger study. If ADOS-2 or recent Mullen scores were unavailable, these assessments were

administered at the end of the session; average time between Mullen administration and T1 was 2.81 months ($SD = 3.60$, Range = 0–11.97), and ADOS administration and T1 was 7.97 months ($SD = 8.17$, Range = 0–31.2). At follow-up (T2; approximately two years following T1), parents completed a second consent form, additional demographic information and questionnaires. All questionnaires were administered online via the Qualtrics survey platform (Qualtrics, 2017).

Data Preparation and Analysis

Missing data analysis revealed that 1.03% (Range 0–7.7%) of data were missing for any single item. Five participants (19.23%) were missing some data (four items on the HSQ-PDD were missing 3.8% of data and four items were missing 7.7% of data). Little's MCAR test indicated data were missing at random, $p = 1.00$. Given pairwise and listwise exclusions would further reduce power for analyses thereby increasing the risk of Type II errors (Newman, 2014), missing values were replaced using multiple imputations in SPSS. Subsequent analyses utilized pooled imputed data. Spearman's rank order correlations were first used to examine the relationships between variables, and were used to inform model entry into hierarchical regressions for further analysis (please refer to Supplementary Materials for additional information regarding assumption checks and additional analyses; <https://www.doi.org/10.26181/5d28087098c9b>).

Results

Demographics

Demographics are provided in Table 1. At baseline (T1), Mullen ELC scores show that, on average, children were developmentally delayed. ADOS-2 CS ranged from 2 to 10 suggesting significant variability in ASD symptom severity between children. At follow-up (T2) children were typically involved in multiple therapies ($M = 3.84$, $SD = 1.90$). The overall sample was characterized as moderately to frequently engaged in challenging behavior on the HSQ-PDD, with these behaviors being rated as occurring at low to moderate severity.

Predictors

Table 2 provides correlations between baseline child characteristics (i.e., developmental level, autistic traits, temperament), and the frequency and severity of challenging behavior. Predictors of frequency and severity of challenging behavior were examined separately.

[Insert Table 2 about here]

Frequency of Challenging Behavior. Frequency of challenging behavior assessed with the HSQ-PDD at follow-up was strongly positively associated with baseline Negative Affectivity ($p < .001$) and negatively with Effortful Control ($p = .024$), but not Surgency-Extraversion ($p = .056$). Additionally, frequency of challenging behavior was not found to be significantly associated with either baseline developmental level assessed with the Mullen ELC ($p = .838$), or with autistic traits assessed with the ADOS-2 ($p = .195$). Next, a linear multiple regression model was applied to examine the unique contributions of Negative Affectivity (T1) and Effortful Control (T1) to Frequency of Challenging Behavior (T2; see Table 3). To control for developmental level, Mullen ELC scores were entered at Step 1 (refer to Table 3) and temperament variables were entered at Step 2. Step 1 accounted for 0.5% of the variance, $F(1, 24) = .119$, $p = .733$. Step 2 accounted for an additional 35.8% of variance, with the full model accounting for 36.3% of variance, $F(3, 22) = 4.19$, $p = .017$; however, only Negative Affectivity significantly and independently predicted the frequency of challenging behavior.

[Insert Table 3 about here]

Severity of Challenging Behavior. Severity of challenging behavior assessed with the HSQ-PDD at follow-up was strongly positively associated with baseline Negative Affectivity ($p < .001$) and negatively with Effortful Control ($p = .001$), Surgency-Extraversion ($p = .001$) and Mullen ELC ($p = .033$). Severity of challenging behavior was not significantly associated with ASD symptom severity on the ADOS-2 ($p = .998$). A linear multiple regression model was applied to examine the unique contributions of Negative Affectivity (T1), Effortful Control (T1), and Surgency-Extraversion to Severity of Challenging Behavior (T2; see Table 4), controlling for developmental level. Mullen ELC scores were entered at Step 1 (refer to Table 4) and temperament variables were entered at Step 2. Step 1 accounted for 31.6% of the variance, $F(1, 24) = 2.61$, $p = .119$. Step 2 accounted for an additional 46% of variance, with the full model accounting for 56% of variance, $F(4, 21) = 6.68$, $p < .001$;

however, none of the temperament variables were found to independently predict the severity of challenging behavior.

[Insert Table 4 about here]

Discussion

The current study examined relationships between baseline ASD symptom severity, developmental delay, and temperament as potential predictors of the frequency and severity of challenging behavior in young children with ASD at 2-year follow-up. To our knowledge, this is the first study to examine the role of temperament in predicting challenging behavior in young children with ASD that is not reliant on cross-sectional methodology. Temperament difficulties associated with effortful control and negative affectivity were significantly correlated with both the frequency and severity of challenging behavior in children with ASD at 2-year follow-up. Additionally, developmental level and surgency-extraversion were found to be associated with severity of challenging behavior. Following further analysis, negative affectivity was identified as a unique predictor of the frequency of challenging behavior. No unique predictors of the severity of challenging behavior were identified, although effortful control approached significance ($p = .063$), and the combined effect of negative affectivity, effortful control, and surgency-extraversion added significant variance to the model predicting severity, over and above developmental level which was not identified as a significant predictor when examined independently.

Our findings tentatively indicate that challenging behavior in young children with ASD may be more likely to be associated with early temperamental difficulties relating to emotional reactivity/dysregulation than developmental level, or even ASD symptom severity. Given the role that emotion regulation plays in creating an “affective homeostasis” or a buffer from which children dynamically and flexibly manage their emotional experience, including everyday stressors (Chow, Ram, Boker, Fujita, & Clore, 2005; Diamond & Aspinwall, 2003), it is not surprising that difficulties in aspects of temperament relating to emotion regulation are predictive of challenging behavior. These findings are consistent with recent studies reporting an association of internal physiological stress with challenging behavior in children and adolescents with ASD (Goodwin, Mazefsky, Ioannidis, Erdogmus, & Siegel, 2019; Nuske et al., 2019), as well as cross-sectional studies comprising older school-aged children/adolescents with ASD that have linked externalizing problems with low effortful control and surgency, and high negative affectivity (Adamek et al., 2011; Burrows et al., 2016; De Pauw et al., 2011; Kamio et al., 2018; Schwartz et al., 2009). Overall, although based on a small number of participants, these results suggest that temperament may be an important characteristic to consider regarding the trajectory of challenging behavior in children with ASD. Importantly, while temperament has been often linked with cognitive, emotional and social development (Kagan & Snidman, 1991), this study fills a notable research gap regarding the role of temperament in the development of challenging behavior in this population.

Unlike Estes et al. (2007) and other cross-sectional studies (e.g. Dominick et al., 2007), our findings failed to identify a strong role for developmental level in the frequency and severity of later challenging behavior in children with ASD, particularly when temperament is considered. The finding that baseline ASD symptom severity was not associated with challenging behavior at follow-up is also inconsistent with some prior research (e.g., Dominick, et al., 2007), possibly reflecting methodological differences across the literature (e.g., our use of the ADOS-2 Comparison Score to assess ASD symptom severity).

Clinical Implications

Targeting temperament difficulties early on in young children with ASD may help to prevent the development of challenging behavior. Given that developmental delay was also associated with severity of challenging behavior (although notably not when including temperament in the model), it may be that if a child who is prone to negative affectivity who is able to harness developmentally appropriate skills and abilities (which broaden their repertoire of coping and problem-solving strategies), the behavior may de-escalate. Conversely, a child with higher negative affectivity who is not able to harness such development skills, including coping and problem solving, may have more severe behavioral outbursts (Kagan, 1997). Therefore, taking into account children’s developmental level, in addition to temperaments difficulties such as effortful control, negative affectivity, or surgency-extraversion, may help to inform intervention programming to prevent or reduce challenging behavior in children with ASD.

Limitations

The study was limited by the small sample size leading to increased risk of Type II error and sample-specific results. Future research with larger samples are required to confirm longitudinal predictions of temperament with challenging behavior in children with ASD. The use of the ECBQ also has its limitations as the factor structure of the instrument has yet to be assessed in children with ASD. While there has been some attention given to the psychometric structure of personality measures in the context ASD (Barger, Campbell, & Simmons, 2014, 2016), the temperament measures often utilized in ASD populations have not yet been subject to such examination (Chetcuti et al., 2019). This would be a useful direction for future work. We note, nonetheless, that the internal reliabilities of the scales reported here offer some support for their use in this population, possibly with the exception of surgency-extraversion due to low Cronbach's alpha. The use of parent report to assess temperament and challenging behavior was also a limitation as results may be affected by parent-report bias (De Los Reyes & Kazdin, 2005). The use of multi-measure methodology (i.e., behavioral, physiological, clinical assessment) is recommended to reduce shared method variance (Chetcuti et al., 2019). Finally, it will be beneficial in future to include a measure of baseline challenging behavior.

Conclusions

Understanding the developmental origins of challenging behavior in children with ASD is critical given the potential impact these behaviors have on the child's cognitive and social development, and on their family's quality of life. Our results identify a potential contribution of early temperamental difficulties on later emerging challenging behavior, particularly dimensions related to emotion regulation. Our findings also suggest a small role for developmental level in the severity of challenging behavior in children with ASD, but suggest this effect may be metered somewhat by temperament.

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Author Contribution

SK, DH, and HN designed the study. SK, DH, LC, ES, and HN collected the baseline data and SK collected the follow-up data. SK and DH analyzed the data. SK, DH, ES and HN drafted the original version of the article, and LC contributed to the revision. All authors critically reviewed the article and approved the final article as submitted.

Conflict of interest

SK declares that she has no conflict of interest. DH declares that he has no conflict of interest. LC declares that she has no conflict of interest. ES declares that she has no conflict of interest. HN declares that she has no conflict of interest.

Compliance with Ethical Standards

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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Table 1
Child Demographics

Demographics	<i>n</i>	%
Gender (T1)	26	100
Male	19	76
Female	7	24
Co-morbid diagnosis (T2): Total ¹	5	19
ASD with GDD	3	12
Genetic conditions (e.g., Fragile X)	2	8
Medications and supplements at T2: Total	10	40
Supplements (e.g., multi-vitamins)	8	32
Psychiatric medication (e.g., Lovan)	1	4
Other (e.g., Ventolin)	1	4
Therapies and interventions (T2): Total	25	96
ABA and behavioral interventions	11	44
Speech pathology	23	92
Occupational therapy	16	64
Psychotherapy	6	24
Social skills	6	24
Other (e.g., play therapy)	5	20
Scales	<i>M</i>	<i>SD</i>
ADOS-2 CS (T1)	7.27	2.31
Mullen ELC (T1)	77.46	26.23
ECBQ (T1)		
Negative Affectivity	3.64	.76
Effortful Control	4.13	.66
Extraversion-Surgency	4.53	.65
HSQ-PDD (T2) ²		
Frequency	16.46	7.00
Severity	2.71	1.86

Note. ¹Co-morbid diagnoses were based off parent report. ²Frequency 0–25, Severity 1–9. ADOS-2 CS: Autism Diagnostic Observation Schedule (2nd edition) Comparison Score. Mullen ELC: Mullen Early Learning Composite (standard score). GDD: Global Developmental Delay. ABA: Applied Behavioral Analysis. ECBQ: Early Childhood Behavior Questionnaire. HSQ-PDD: Home Situations Questionnaire-Pervasive Developmental Delays.

Table 2
Spearman’s Rank Order Correlations between Child Characteristics (T1) and Challenging Behavior (T2), *n* = 26

	1.	2.	3.	4.	5.	6.
1. Mullen ELC (T1)	-					
2. ADOS-2 CS (T1)	-.238	-				
3. ECBQ Negative Affectivity (T1)	-.234	-.253	-			
4. ECBQ Effortful Control (T1)	.238	-.008	-.574**			
5. ECBQ Surgency-Extraversion (T1)	.459*	.063	-.563**	.447*		
6. HSQ-PDD Frequency (T2)	-.043	-.264	.637***	-.438*	-.378	
7. HSQ-PDD Severity (T2)	-.417*	-.001	.641***	-.581**	-.587**	.715***

Note. Mullen ELC: Mullen Early Learning Composite (standard score). ADOS-2: Autism Diagnostic Observation Schedule (2nd edition) Comparison Score. ECBQ: Early Childhood Behavior Questionnaire. HSQ-PDD: Home Situations Questionnaire, Pervasive Developmental Delay.

p* < .05 *p* < .01 ****p* < .001

Table 3
Linear Regression Model of Predictors of the Frequency of Challenging Behavior

	<i>b</i>	<i>SEB</i>	<i>β</i>	<i>p</i>
Step 1				
Constant	15.01	4.43	–	.001
Mullen ELC	.019	.054	.070	.730
Step 2				
Constant	5.06	14.15	–	.721
Mullen ELC	.04	.047	.150	.394
ECBQ Negative Affectivity	4.46	1.89	.485	.019
ECBQ Effortful Control	–1.92	2.26	–.180	.397

Note. Step 1, $R^2 = .005$, $p = .773$; Step 2, $R^2 \Delta = .358$, $p = .007$. Mullen ELC: Mullen Early Learning Composite. ECBQ: Early Childhood Behavior Questionnaire.

Table 4

Linear Regression Model of Predictors of the Severity of Challenging Behavior

	<i>b</i>	<i>SEB</i>	β	<i>p</i>
Step 1				
Constant	4.52	1.11	–	<.001
Mullen ELC	–.023	.014	–.313	.099
Step 2				
Constant	7.59	4.18	–	.069
Mullen ELC	–.009	.012	–.105	.455
ECBQ Negative Affectivity	.717	.478	.291	.134
ECBQ Effortful Control	–.945	.508	–.325	.063
ECBQ Surgency-Extraversion	–.644	.544	–.261	.236

Note. Step 1, $R^2 = .100$, $p = .116$; Step 2, $R^2 \Delta = .460$, $p = .002$. Mullen ELC: Mullen Early Learning Composite. ECBQ: Early Childhood Behavior Questionnaire.

Supplementary Material

[Click here to download e-component: Supplementary.pdf](#)

Highlights

- Temperament, development and ASD symptom severity were examined as predictors of challenging behavior
- Negative affectivity predicted frequency of challenging behavior at follow-up
- No individual variables uniquely predicted severity of challenging behavior at follow-up
- Emotion regulation-related aspects of temperament may predict later challenging behavior in ASD