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# Exploring Early Temperament Predictors of Maladaptive Behavior in a Group of Elementary School Age Children with Autism Spectrum Disorder

### Abstract

The current study is an *ex post facto* nonexperimental design analyzing archival data collected from previous trials of a large-scale longitudinal study conducted by researchers at JFK Partners, Center of Excellence in Autism and Neurodevelopmental Disabilities in collaboration with the University of Colorado Anschutz Medical School (Principal Investigator: Susan Hepburn, Ph.D.). Specifically, the study looked at the temperament characteristics of mood, intensity, adaptability, and approach measured within early childhood and how they relate to later maladaptive behavior within a group of elementary age children diagnosed with ASD. Maladaptive behavior is a particularly salient outcome, as it impacts the child and family above and beyond the challenges associated with ASD (Hartley, Sikora, and McCoy, 2008; Hastings et al., 2005; Lecavalier, Leone, & Wiltz 2006). There is an established body of literature exploring how temperament can act as a risk factor that predisposes individuals who are typically developing to a variety of later negative outcomes, including maladaptive behavior (Belsky & Pluess, 2009; Boyce, 2016; Earls & Jung, 1987; Martin, 1994; Rothbart, 2012a; Thomas & Chess, 1977). However, more research is needed to explore temperament characteristics and later maladaptive behavior for children with ASD.

A sample of 71 children with ASD met the inclusion criteria within the archival longitudinal dataset. Power analyses indicated that the results were underpowered and thus should be interpreted with caution. Results yielded statistically significant correlations between approach, intensity, and mood with later maladaptive behavior. Further, when controlling for estimated cognitive ability, gender, and maternal education, significant predictive relationships were found between the temperament characteristics of mood, intensity, and approach with later maladaptive behavior. Finally, the combined temperament characteristics yielded significant results when predicting later maladaptive behavior. Notably, the temperament construct of adaptability was not significantly related to or predictive of later maladaptive behavior within the current study.

The results highlighted the construct of mood, and related constructs, as particularly salient predictors of later maladaptive behavioral outcomes within children with ASD. Additionally, intensity and approach were predictive of externalizing and internalizing behavior respectively. Looking at the combination of temperament factors may also have important implications regarding later behavioral outcomes with children with ASD. Thus, the results highlight that examining temperament as a risk factor for the development of later maladaptive behavior for children with ASD is an important area of study that requires future inquiry.

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of the Requirements for the Degree

Doctor of Philosophy

by

Caren D. Rhodes-Doudna

June 2019

Advisor: Cynthia Hazel, Ph.D.

Copyright by Caren Rhodes-Doudna 2019 All Rights Reserved Author: Caren Daneka Rhodes-Doudna Title: Exploring Early Temperament Predictors of Maladaptive Behavior in a Group of Elementary School Age Children with Autism Spectrum Disorder Advisor: Cynthia Hazel, Ph.D. Degree Date: June 2019

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#### **Chapter One: Introduction and Study Purpose**

Autism Spectrum Disorder (ASD) is a complex disorder that impacts individuals' social/communication, behaviors, and thought processes throughout their lifespan. While individuals with ASD can have a wide range of functioning, the diagnosis of ASD can cause an enormous amount of stress for an individual and their family, and thus can impact the quality of life for an individual and their family. The significant impact of a diagnosis of ASD can be thought of systemically from an individual, family, community and policy perspective (Karst & Hecke, 2012).

A primary source for ASD prevalence statistics within the United States is the Center of Disease Control and Prevention (CDC). The statistics are released twice per year, based on a review of medical and educational records (when available) of children who are eight years old from eleven sites that have agreed to ASD monitoring from four years before the reported statistics (e.g. the prevalence statistics reported in 2018 are from 2014 records) (Baio et. al, 2018). The CDC indicated that the current prevalence of Autism Spectrum Disorder (ASD) within the United States is 1 in 59. This is a 15% increase from previously reported prevalence statistics of 1 in 68 (see Figure 1; Autism Speaks, 2018). Further, individuals identified as male are four times more likely to receive a diagnosis of ASD than those identified as females. ASD prevalence information is reported consistently across all racial, ethnic, and socioeconomic groups (Autism Speaks, 2018; Baio et.al, 2018). Additionally, according to the World Health Organization (2017), the estimated average global incidence of ASD is 1 in 160 children and there is general agreement that the prevalence of ASD is increasing worldwide (World Health Organization, 2017). While there is no consistent or concise explanation for the steady increase in ASD, there is debate as to whether the actual incidence of ASD is increasing or if the processes and procedures of surveillance and identification have become more refined, and thus more accurate and readily available (World Health Organization, 2017; Wright, 2017).

Thus, there is a critical need for more research on what influences outcomes for individuals with ASD as well as how to provide them effective supports. A deeper understanding of factors that influence outcomes will better prepare clinical and educational professionals to provide interventions and work towards prevention.



*Figure 1.* 2018 Autism Prevalence Estimate Based on the image from Autism Speaks, 2018 (<u>https://www.autismspeaks.org/science-news/cdc-increases-estimate-autisms-prevalence-15-percent-1-59-children</u>)

#### **Overview of ASD**

ASD is a pervasive disorder that impacts individuals' fundamental abilities to communicate, relate, and interact socially across each stage of development. Individuals with ASD also must have a broad category of symptoms called rigid and repetitive behaviors (American Psychiatric Association, 2013; Kim et al., 2014). Examples of rigid and repetitive behaviors are ritualistic, perseveration, compulsive, and/or obsessive behavior (Watt, Wetherby, Barber & Morgan, 2008). Individuals with ASD have a wide range of functioning and the core characteristics have a unique presentation for each individual with the diagnosis. The symptoms of ASD can be apparent early in development; however, the characteristics may not fully manifest until the demands of the individual's social environment exceed their ability to function in a healthy way (American Psychiatric Association, 2013; Happé, Ronald, & Plomin, 2006; Zaky, 2017;). ASD commonly co-occurs with a variety of conditions including, but not limited to, intellectual disability, anxiety, depression, attention deficit and hyperactivity disorder, mood disorder, tic disorders, and seizure disorders, which compound the level of stress and functional impact for individuals and their families (Cadman et al., 2012; Kring, Greenberg, & Seltzer, 2008; Yorke et al., 2018). Further, ASD is a neurological condition, and research suggests that there are physical differences within the structure of a brain of individuals with ASD when compared to individuals without ASD (Happé, Ronald, & Plomin, 2006). Researchers and scientists largely agree that there is no single cause of ASD; however, there is likely a genetic component to the etiology of the disorder.

#### **Early Intervention**

Notably, there is no known 'cure' for ASD. However, it is a condition that often responds to intervention, particularly early intervention. In order to have more positive future outcomes, individuals with ASD often need to be directly taught social skills, coping strategies, functional communication, and other skills related to self-regulation that individuals who are neurotypical learn more automatically (Lord, Elsabbagh, Baird, Veenstra-Vanderweele, 2018). Once a child is diagnosed with ASD, it is critical that a comprehensive treatment plan is promptly developed to support the child at home, in the community, and at school. Often individuals who receive a diagnosis of ASD have multiple areas of need and it can be difficult to know what areas to prioritize within intervention (Bailey et al., 2006). Other components, like shock or grief, after receiving a diagnosis can make the diagnostic process overwhelming for families. It can be difficult to make intervention decisions and navigate systems when trying to cope with these strong emotions. Gaining clarity regarding what areas of need to emphasize and prioritize within intervention planning is beneficial for families and professionals as a next step after the diagnostic process (Bailey et al., 2006).

#### **Conceptual Models Explaining Outcomes**

Prevention science suggests that if risk factors are identified, predictions can be made about potential stressors and protective factors that inform intervention and treatment (Coie et al., 1993). Within the field of development, models like the diathesisstress and dual risk models suggest that individuals have genetic vulnerability that predisposes them reacting more negatively to stress and thus, ultimately have broader negative outcomes (Belsky & Pluess, 2009). However, developmental models of child outcomes emphasize the interactions between individuals and their environments over time. Individual child factors combined with environmental factors and their interactions have the potential to significantly and uniquely impact a child's development over time (Belsky, Bakermans-Kranenburg, & van IJzendoorn, 2007; Belsky & Pluess, 2009; Brofenbrenner, 1979).

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#### **Differential Susceptibility Models**

Individuals vary greatly in their responses to stress and their exposure to social and environmental stressors (Boyce, 2016). A primary focus of developmental and evolutionary theorists has been the stress response system of an individual when considering resiliency and outcomes (Boyce, 2016; Ellis, Essex, & Boyce, 2005). Researchers have elaborated on concepts of the diathesis stress model of vulnerability and empirically supported theoretical models suggest that an individual may be particularly susceptible to both the positive and the negative effects of their environment (Boyce, 2016; Giudice, 2016). Models of differential susceptibility integrate developmental, evolutionary, and genetic points of view and suggest that there are complex interactions between the quality of the environment experienced by an individual and their fundamental neurodevelopmental/biological traits (e.g. temperament) (Belsky & Pluess, 2009). The two main theories that incorporate this concept are Differential Susceptibility Theory (DST) proposed by Belsky and colleagues (1997) and Biological Sensitivity to Context (BSC) proposed by Boyce and Ellis (2005). Both theories argue that there are individual differences in an individual's susceptibility to contextual factors within their life (Belsky & Pluess, 2009). Inherently vulnerable individuals may also benefit the most from environmental supports or simply the absence of adversity (Ellis & Boyce, 2008). Further, both the BSC and DST theories are based on an evolutionary philosophy, but they put a different emphasis on what constitutes developmental plasticity across environments (Belsky & Pluess, 2009).

#### **Biological Sensitivity to Context (BSC).**

The metaphor. Metaphors can be particularly helpful when explaining and conceptualizing complex ideas (Kendall-Taylor & Haydon, 2016). In the field of development there is a metaphor that Boyce and Ellis (2005) created using an evolutionary perspective which compares children to either orchids or dandelions when considering what contributes to resilience and outcomes. Specifically, 'orchid children' describe children who are highly sensitive to the positive and negative characteristics of the environment. Children who fall into the orchid group are particularly sensitive to adverse experiences. Children who Boyce and Ellis (2005) compare to dandelions are not as susceptible to environmental characteristics and stimuli. These children generally demonstrate resilient characteristics regardless of what is happening around them, and thus have more positive outcomes (Boyce & Ellis, 2005; Ellis & Boyce, 2008; Kendall-Taylor & Hayden, 2016). Recently, Lionetti and her colleagues (2018) further developed the metaphor and suggested that there is a third group of individuals who are of medium sensitivity to environmental stimuli that labeled 'tulips.' Children within this group are not as sensitive as children within the orchid group, but more sensitive than those within the dandelion group. The idea that children have different degrees of susceptibility to their environment and experiences puts an emphasis on how specific factors, in this case individuals' genetic predisposition, can influence a child's ability to process and learn from their experiences and ultimately, their outcomes and ability to thrive (Boyce, 2016; Boyce & Ellis, 2005). Individual differences in child characteristics like temperament, cognitive ability, gender, and other neurobiological factors can strongly influence how

children process their environment and experiences as well how they respond to adversity (Boyce, 2016; Ellis & Boyce, 2008; Lengua & Wachs, 2012). The current study seeks to understand more about individual risk factors and determine if some factors are more salient than others when predicting negative outcomes for children with ASD.

The Theory. The BSC model has its' foundations in developmental research on child health and negative outcomes. Specifically, Boyce and colleagues (1995) originally predicted outcomes for children with cardiovascular and immune reactivity. They found when exposed to positive/supportive environments, children; however, they had worse outcomes when exposed to stressful environments (Boyce et. al, 1995). Thus, the researchers used a differential susceptibility hypothesis to explain their research results. Boyce and Ellis (2005) later elaborated on their hypothesis and proposed the BSC model, in which they purported that an individual's sensitivity to their environment (or their "context") is primarily determined by individual differences within neurobiological characteristics of an individual. Further, the biological sensitivity-to-context model states that the stress-response system is a conditional response that starts from an early age to modify itself based on environmental conditions and life experience (Boyce & Ellis, 2005). Specifically, Boyce and Ellis (2005) indicated that a heightened plasticity to environment is adaptive within harsh, stressful environments or within positive environments with protective factors. Plasticity or sensitivity within a stressful environment, according to the BSC, increases an individual's ability to detect and respond to dangers and threats. In protective environments, plasticity helps an individual maximize the benefit they receive from resources and support (Boyce & Ellis 2005).

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Notably, despite the proposed benefits of plasticity, Boyce and Ellis (2005) emphasized that even in environments that are moderately stressful, the evolutionary cost of being highly susceptible to the environment ultimately outweighs the benefits (Boyce, 2016; Boyce & Ellis, 2005).

**Differential Susceptibility Hypothesis**. The differential susceptibility hypothesis also suggests that there is an evolutionary basis for how susceptible a child is to contextual or environmental factors within their life, and this can impact outcomes (Belsky, 1997, 2005; Belsky & Pluess, 2009). Similarly, Belsky (1997) emphasized that an individual's plasticity is largely based individual characteristics that are genetically determined. Specifically, individuals who are less sensitive to their environment are less vulnerable to the stresses that they experience. This can also be protective for individuals who may have genetically based characteristics that make them particularly vulnerable to the environment. Later research conducted by Ellis and colleagues (2011) suggested that specific individual factors like temperamental reactivity are likely mediators of this plasticity (Ellis, Boyce, Belsky, Bakermans-Kranenburg, & Ijzendoorn, 2011). Over time, the DST model evolved to become more technical and now includes at minimum two interactions with the environment that then determines positive or negative outcomes. The first interaction is the gene with environment interaction (GxE), which determines a child's plasticity. Subsequently, the child's level of developmental plasticity relative to the environmental factors that they experience (PxE) is purported to determine potential outcomes (Guidice, 2016).

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#### Temperament

Thomas, Chess, and Birch (1968) conceptualized temperament at the behavioral style of the individual. While there are other definitions of temperament that will be explored within the literature review, this will be the definition also used within the current study. Temperament plays a key role in how individuals respond to novel situations and adversity (Lengua & Wachs, 2012). There is some literature to suggest that temperament can be measured as early as when a child is a few months old (e.g. reactivity) and should be fully apparent by the preschool years (Zentner & Bates, 2008). At the time of Thomas and Chess' (1957) foundational New York Longitudinal Study (NYLS), there was a strong push for the exploration of the interaction of environmental factors with child characteristics. The NYLS followed over 100 individuals, primarily from educated families living in New York, from the age of three months to adulthood, measuring temperament using a parent interview approach (Thomas & Chess, 1986). The NYLS set the stage for temperament being an important, relevant predictor of later child outcomes. The results of the NYLS, as well as a multitude of subsequent studies, established that certain temperament characteristics can be considered as risk factors (e.g. negative mood, high intensity, low adaptability, and low approach), which can predispose an individual to, and thus predict, negative outcomes like maladaptive behavior, poor physical and social/emotional health, academic difficulty, and an overall lower quality of life (Belsky & Pluess, 2009; Earls & Jung, 1987; Martin, 1994; Rothbart, 2012a; Thomas & Chess, 1977). Over the past decades, the knowledge base regarding individual differences in temperament, how important these differences are within a variety of

contexts, and ways to address the behavioral and functional outcomes that may result from these differences have grown exponentially (Rothbart, 2012b). However, more information is needed on risk factors like temperament and how temperament characteristics interact with each other to influence the outcomes of individuals (Fowles & Dindo, 2009). There is a particular need to research if the relationships within the existing literature regarding risk factors and outcomes hold true for individuals who are identified with ASD.

#### **Problem Statement**

With the identification of ASD increasing more research is needed to understand what influences outcomes for children within this population (Levy & Perry, 2011). More research can allow for more effective intervention and prevention strategies for the parents, educators, and clinicians interact with children with ASD every day. Additionally, more evidence-based intervention and prevention strategies can improve outcomes for children with ASD and their families. Due to the core deficits inherent in the diagnosis of ASD, there is a broad body of literature that argues that children with ASD are more likely to demonstrate maladaptive behavior, both externalizing and internalizing, than children who are typically developing and those who have intellectual disability without co-occurring conditions (Hartley, Sikora, and McCoy, 2008). The specific skill deficits under the broader core deficits of ASD can ultimately lead to a wide array of internalizing and externalizing behavior that impact an individual with ASD's long-term ability to progress, learn, develop, and function independently (Sharma, Gonda, & Tarazi, 2018). Maladaptive behavior is a particularly salient negative outcome that can often lead to significant functional impairment both for the individual child within their home, school, and community as well as for the quality of life of the family unit. Further, maladaptive behaviors almost always have a significant functional impact on the child and family and often cause caregiver distress above and beyond the core symptoms of ASD (Hartley, Sikora, and McCoy, 2008; Hastings et al., 2005; Lecavalier, Leone, & Wiltz 2006). Additionally, the lagging skills that often underlie maladaptive behavior impact overall quality of life and emotional well-being (Green, 2008; Maddox et al., 2018).

Thus, given how common chronic maladaptive behavior is within individuals with ASD throughout their lifespan, and how pervasive the negative impact of these behaviors can be, it is critical that we learn more about what predicts maladaptive behavior for children with ASD (Weiss, Cappadocia, MacMullin, Viecili, & Lunsky, 2012). Exploring the connections between individual factors like temperament and maladaptive behavior can provide valuable information on predictors of maladaptive behavior and can improve options for intervention and prevention for children with ASD.

#### **Goals of the Current Study**

There were two broad, primary goals of the current study. The first is to provide information to families, practitioners, and educators to guide expectations for prognosis of ASD and to aid in the intervention and prevention planning process. Providing more information about the individual characteristics of children with ASD may help prevent and/or mitigate negative outcomes for children and families. Secondly, there are wellestablished relationships between individual risk factors, like temperament, and the development of maladaptive behavior within individuals who are typically developing (Belsky & Pluess, 2009; Boyce, 2016; Earls & Jung, 1987; Martin, 1994; Rothbart, 2012a; Thomas & Chess, 1977). The current study sought to explore if these established relationships are the same or different for individuals with ASD. The existing research indicates that children with ASD are at a greater risk for developing maladaptive behavior. Thus, gaining a deeper understanding about what specific factor or combination factors are most predictive of outcomes would enhance the existing literature.

Anticipatory Guidance. Anticipatory guidance is a term often used within the medical literature and can be defined as providing therapeutic, preventative advice based on known child characteristics (Schuster, Duan, Regalado, & Klein, 2000). The foundations of present- day temperament research originated when pediatricians Carey and McDevitt (1986) wanted to give parents temperament-based anticipatory guidance regarding what to expect as they raised their children. There is a vast body of literature that exists in the field of temperament focusing on child and family outcomes, the stability of temperament, and the inheritability of temperamental traits (Rothbart, 2012a). In previous studies, parents reported that when they received temperament-based anticipatory guidance, that was temperament-based they felt more prepared for maladaptive behavior and were able to provide more specific intervention. Parents who felt more prepared to prevent or address their child's behavior based on their

temperament reported improvement in the parent-child relationship (Cameron, Rice, Sparkman, & Neville, 2013).

Therefore, given that temperament has been established as a valuable contributor to outcomes within the typically developing population, more information is needed on how best to provide effective anticipatory guidance based on known temperament characteristics for children with ASD (Camarata, 2014). With this information, treatment teams may be able to give families better, more individualized guidance after their child has received an ASD diagnosis. Additionally, if individual temperament factors, or a combination of factors, are identified as predictive of maladaptive behavior, then families and intervention teams may be able to prioritize specific aspects of their intervention planning in order to target specific needed skills (e.g. emotional regulation, cognitive flexibility) for prevention or mitigation of maladaptive behavior

**Understanding the Impact of Individual Risk Factors**. Another primary goal of the current study is to specifically explore the associations between early temperament characteristics, specifically mood, approach, intensity, and adaptability of children with ASD, and later maladaptive behavior within a sample of children diagnosed with ASD. Ultimately, understanding the nuances of how individual risk factors influence outcomes for children with ASD adds valuable, needed information to the field. If the relationships of these risk factors differ from the typically developing population, it will be important to explore and describe these differences and what the implications are for intervention, prevention, and research. If the relationships are consistent with the typically developing

population, it will be important to individualize the results to fit the unique needs of children with ASD and their families.

#### **Research Questions**

**Question One.** What specific temperament characteristics in early childhood are related to overall, internalizing, and externalizing maladaptive behavior in later childhood in a sample of children with Autism Spectrum Disorder (ASD)?

*Hypothesis One.* There is a statistically significant correlation between the temperament subcategories of: adaptability, intensity, mood, and approach identified in early childhood and overall, internalizing, and externalizing maladaptive behavior later in childhood in a group of children identified with ASD.

**Question Two**. After accounting for maternal education, gender, and cognitive functioning, do the specific temperament characteristics of adaptability, intensity, approach, and mood significantly predict overall, internalizing and externalizing maladaptive behavior later in childhood in a group of children with ASD?

In general, it is hypothesized that early temperament characteristics significantly predict maladaptive behavior later in childhood above and beyond maternal education, gender, and cognitive functioning in children with ASD. Specifically:

*Hypothesis 1.* Controlling for maternal education, gender, and cognitive functioning, overall, internalizing, and externalizing maladaptive behavior in later childhood in children with ASD is statistically significantly predicted by the temperament characteristic of adaptability.

*Hypothesis 2.* Controlling for maternal education, gender, and cognitive functioning, overall, internalizing, and externalizing maladaptive behavior in later childhood in children with ASD is statistically significantly predicted by the temperament characteristic of intensity.

*Hypothesis 3.* Controlling for maternal education, gender, and cognitive functioning, overall, internalizing, and externalizing maladaptive behavior in later childhood in children with ASD is statistically significantly predicted by the temperament characteristic of mood.

*Hypothesis 4.* Controlling for maternal education, gender, and cognitive functioning, overall, internalizing, and externalizing maladaptive behavior in later childhood in children with ASD is statistically significantly predicted by the temperament characteristic of approach.

*Hypothesis 5.* Controlling for maternal education, gender, and cognitive functioning, overall, internalizing, and externalizing maladaptive behavior later in childhood in children with ASD is statistically significantly predicted by the combination of temperament characteristics of adaptability, intensity, mood, and approach

#### **Definitions.**

Adaptability. Adaptability is a temperament characteristic that reflects how quickly and easily an individual adjusts positively or negatively to environmental changes or novelty within the environment (Carey, 1998; Nelson, Martin, Hodge, & Kamphaus, 1999). **Approach/Withdrawal**. Approach is a temperament characteristic referring to the initial response a person has to new stimuli (Thomas, Chess, & Birch, 1970).

**Differential Susceptibility Models.** Models of differential susceptibility integrate developmental, evolutionary, and genetic points of view and suggest that there are complex interactions between the quality of the environment that the individual experiences and their fundamental neurodevelopmental/biological traits (e.g. temperament) (Belsky & Pluess, 2009; Boyce, 2016).

"Difficult" versus "Highly Sensitive" Temperament. Children characterized as having a 'difficult temperament' are defined in the literature as having a temperament profile that includes high intensity reactions, low adaptability and approach, and general negative mood (Thomas & Chess, 1986). Within the current study, consistent with the language used within the differential susceptibility theories of Belsky and Pluess (2009) as well as Boyce and Ellis (2005), "highly sensitive" temperament will be used to describe "difficult temperament."

**"Easy" Temperament**. Children having the temperament characteristics of positive mood, low/moderate intensity of reaction, and high adaptability (Thomas & Chess, 1986).

**Externalizing Behavior**. "Undercontrolled" (Quay & La Greca, 1986); Externalizing behavior is a broad term that includes concepts like aggression, disruptive behavior, antisocial behavior, impulsivity and conduct problems (Rothbart, 2012a; Hinshaw, Han, Erhardt & Huber, 1992; Silver, Measelle, Armstrong, & Essex, 2005). **Intensity.** A temperament characteristic that refers to the energy and intensity of the emotional reaction to a stimulus. Thomas and Chess (1977) define intensity as the energy level of responses "regardless of quality or direction," (Carey, 1998, p.524).

**Internalizing Behavior**. "Overcontrolled" (Quay, 1986); Internalizing behavior is a broad term used to describe behaviors such as anxiety, depression/dysphoria, and withdrawal (Rothbart, 2012b; Hinshaw, Han, Erhardt, & Huber, 1992).

**Mood.** Mood is a temperament characteristic referring to the degree of pleasant or friendly behavior in response to environmental stimuli (Thomas, Chess, & Birch, 1970).

**Problem Behavior versus Maladaptive behavior**. These terms are synonymous in the literature; however, the term "maladaptive behavior" will be used within the current study. The term maladaptive behavior is more objective and has less of a value judgement attached to it. Note that within the literature "problem behavior" is still a term that is widely accepted and used. Maladaptive behaviors are behaviors that have a negative functional impact for the child and family.

**Temperament.** In the current study, temperament will be defined as the behavioral style of the individual (Buss & Plomin, 1986; Mervielde & De Pauw, 2012). A helpful distinction between temperament and behavior is that temperament represents the "style and form," the nature, or the "how" of the behavior (Krieger & Stringaris, 2015, p.2). Further, temperament, as the *how* of behavior, is distinct from the *why* of the behavior (the function) and the *what* of the behavior, or the talents or perceptions of an individual (Chess, 1990).

#### **Chapter Two: Review of the Literature**

#### Introduction

The following literature review focuses on temperament, maladaptive behavior, the known relationships between temperament and maladaptive behavior, and the gaps in the literature regarding temperament and maladaptive behavior for children with Autism Spectrum Disorder (ASD). The focus of this literature review will be to give the reader general contextual information regarding the concepts within the study and then to elaborate on specific information relevant to the study.

#### **Children with Autism Spectrum Disorder**

Autism Spectrum Disorder (ASD) is a pervasive neurodevelopmental disorder that impacts individuals' fundamental abilities to communicate, relate, and interact socially across each stage of development. Additionally, individuals with ASD often have a broad category of characteristics called rigid and repetitive behaviors (American Psychiatric Association, 2013; Kim et al., 2014). Per a 2018 Center of Disease Control (CDC) report, within the United States, the prevalence of ASD is 1 in 59, with males being four times more likely to be identified with ASD than females (Baio et.al, 2018). While researchers and scientists largely agree that there is no single cause of ASD, there is a genetic component to the etiology of the disorder (Abrahams & Geschwind, 2008; Geschwind, 2009). Individuals with ASD have a wide range of functioning and the core characteristics have a unique presentation for each individual with the diagnosis. The symptoms of ASD can be apparent early in development; however, the characteristics may not fully manifest themselves until the demands of the individual's social environment exceed their ability to function in a healthy way (Happé, Ronald, & Plomin, 2006; Zaky, 2017). ASD is a neurological condition, and research suggests that there are physical differences within the structure of a brain of individuals with ASD when compared to individuals who do not have ASD (Geschwind, 2009; Happé, Ronald, & Plomin, 2006). Notably, while there is no known 'cure' for ASD, it is a condition that responds to early, intensive, structured intervention. Specifically, in order to have more positive future outcomes, individuals with ASD often need to be directly taught social skills, coping strategies, functional communication, and other skills related to self-regulation that individuals who are neurotypical learn more naturally (Chaby, Chetouani, Plaza, & Cohen, 2012).

The Historical Context of Autism Spectrum Disorder. Autism comes from the Greek word "autos," which means self (Kita & Hosokawa, 2011). Notably, well before the word autism was recognized by researchers and the professional community, there were several documented cases of individuals who likely had ASD. One of the earliest, and most famous, documented cases was "The wild boy of Averyon" in France in 1799, about a boy named Victor (Donvan & Zucker, 2016; Woody & Viney, 2017). Physician Philippe Pinel worked with Victor who had been abandoned by his parents but survived alone in the woods presumably for years. As a result, he had extreme delays in all areas of his development. After care and intervention by Pinel, Victor's receptive language

and adaptive skills improved, but never was able to acquire verbal language (Woody & Viney, 2017).

The term 'autism' was first used in 1911 by Eugen Bleuler when referring to individuals in adulthood or early adolescence with schizophrenia who were demonstrating difficulty with reciprocal communication and who were avoiding social interaction and isolating themselves (Kita & Hosokawa, 2011; Zaky, 2017). However, it was not until 1943 that psychiatrist Leo Kanner at John's Hopkins Hospital used the term infantile autism to refer to a pediatric population with social/emotional deficits that mirror what is currently conceptualize as ASD (Kita & Hosokawa, 2011). Kanner's most notable work is called the Autistic Disturbances of Affective Contact, based on the observations of eleven children he worked with that exhibited symptoms consistent with lower functioning ASD. Within his small sample, Kanner noted language atypicalities, difficulty with social interaction/contact, and a strong memory for specific information (Kita & Hosokawa, 2011). Further, Kanner noted that the children had difficulty learning adaptive skills, mood lability, and an "insistence on sameness," (Kita & Hosokawa, 2011, p.149). Additionally, in Germany around 1944, Hans Asperger published an article in German using the word 'autistic' to describe four children who had very specific, strong memories, social skills deficits, and narrow interests (Kita & Hosokawa, 2011). While Asperger reported some language abnormalities in these four case studies, he reported higher cognitive abilities and less severe language delays than Kanner reported. In general, the individuals that Asperger described were higher functioning (Kita & Hosokawa, 2011).

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As the field of psychology evolved so did the definition, conceptualization, and treatment of ASD. For example, in the 1950s and 60s, the etiology of ASD was suggested to be pathology that had its foundation within parents, particularly mothers (commonly referred to as "Refrigerator Mothers") (Jack, 2014; Sousa, 2011). Additionally, at that time ASD was conceptualized as being rare and related to schizophrenia (Baker, 2013; Laidler, 2005; Zaky, 2017). In the 1970s, thoughts regarding ASD's etiology shifted to having a biological origin within the individual and continued to be considered low incidence. ASD was seen as a psychiatric disorder separate from schizophrenia, treated like many psychiatric conditions at the time through LSD, electro convulsive therapy, and negative punishment (Baker 2013; Zaky, 2017). Further, when the Diagnostic Statistical Manual, Third Edition (DSM-III) was published in 1980, Infantile Autism continued to be defined as separate from Schizophrenia and was considered to have its own characteristics and definitions. In the Diagnostic Statistical Manual, Third Edition, Revised (DSM-III-R), 'Autistic Disorder' was defined as separate from Infantile Autism (Kita & Hosokawa, 2011). In 1981, Dr. Lorna Wing published an article that disseminated the knowledge of 'Asperger disorder' as conceptually separate from Autism internationally (Kita & Hosokawa, 2011). It took years for researchers, clinicians, and the community to differentiate Autism and Asperger Disorder and to understand the differences between the two conditions. In 1994, the DSM-IV and DSM-IV-TR were published describing differences between Autistic Disorder and Asperger Disorder under the broader category of Pervasive Developmental Disorders (Kita & Hosokawa, 2011). Other disorders under this broad umbrella of

Pervasive Developmental Disorders within these manuals included Rett's disorder, Childhood Disintegrative Disorder, and Pervasive Developmental Disorder Not Otherwise Specified, (PDD-NOS), (Kita & Hosokawa, 2011).

**Current Definition of ASD**. The current definition of Autism Spectrum Disorder (ASD) is found within the Diagnostic Statistical Manual, Fifth Edition (DSM-5), which was released in May of 2013 (Kim et al., 2014). The DSM-5 is the current manual used by clinicians and researchers to diagnose psychological disorders. Diagnoses like Asperger syndrome and Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS) have both been subsumed under the broad diagnosis of ASD within the DSM-5. There is also a new related diagnosis that can be found within the DSM-5 called Social Communication Disorder (SCD) (Kim et al., 2014). Research suggests that most individuals with previous diagnoses of Asperger Disorder, Autistic Disorder, and PDD-NOS meet the DSM-5 diagnostic criteria for either ASD or SCD (Kim et al., 2014). Further, the DSM-5 takes a dimensional approach to diagnosis as opposed to the categorical approach which was used in previous versions of the manual. This was a major paradigm shift within the conceptualization of diagnosis within the field of psychology (Kim et al., 2014). The dimensional approach emphasizes a continuum-based assessment versus isolated categories and meeting specific thresholds in order to be assigned a diagnosis (Kim et al., 2014). Further, once an individual receives an ASD diagnosis, the DSM-5 has levels of severity to clarify the impact of the disorder, ranging from Level 1, requiring support in the areas of social/communication and/or restricted interests and repetitive behaviors to Level 3, requiring very substantial support in the
areas of social communication and/or restricted interests and behaviors (American Psychiatric Association, 2013). Notably, it is important to recognize that these severity levels are not used to influence the level of services needed for an individual nor should they be used for eligibility or qualification of services (American Psychiatric Association, 2013).

Fundamental Characteristics of Autism Spectrum Disorder. Specifically, the core deficits associated with ASD are difficulty with reciprocity within their social/emotional skills, deficits within social nonverbal communication, difficulty understanding relationships as well as developing and maintaining them, and restrictive and/or repetitive patterns of behaviors, interests, or activities (American Psychiatric Association, 2013). The core symptoms are present within early development and can cause significant difficulty within functional skills in a variety of areas of their life throughout their lifespan (American Psychiatric Association, 2013). Multiple retrospective home video studies of individuals later diagnosed with ASD yielded differences in social/emotional development in infancy; however, there is dispute in the literature regarding how early signs of ASD can occur and presentation may vary based on the individual's level of functioning (Matson, Wilkins, & Gonzalez, 2008; Osterling & Dawson, 1994). Examples of early characteristics of ASD can include, but are not limited to, a reactive temperament, difficulty with eye contact, decreased emotional affect, communication delays, delays in joint attention, cognitive inflexibility, other nonverbal communication delays, and delays in pretend play (Osterling & Dawson, 1994). As children develop, their social and communication deficits can be more

apparent as the demands of the social environment increase. Notably, the deficits may be present in early development, but the functional impact may not fully be seen until the developmental social and communication demands supersede the individual's ability to adapt, cope, and/or access and benefit from the resources available to them (American Psychiatric Association, 2013).

**DSM-5 Diagnostic Criteria**. It is important to note, that while individuals with ASD have the same core deficits, the heterogeneity (or variability) with which deficits manifest is significant and there is a wide range of functional impact that ASD has on each individual (Georgiades, Szatmari, & Boyle, 2013; Jeste & Geschwind, 2014; Lenroot & Yeung, 2013). For example, children with ASD can range from having an intellectual disability and limited verbal capabilities, to highly developed cognitive abilities and hyperlexic capabilities. (Georgiades, Szatmari, & Boyle, 2013; Jeste & Geschwind, 2014). The nuances and complexity of ASD require a high degree of specialization to accurately identify and treat. It can take years, particularly in high functioning individuals, for parents to receive an accurate diagnosis for their child (Geschwind, 2009).

It is critical for professionals, and ideally a team of professionals, to be familiar with conditions such as intellectual disability, other global developmental delays, and different disorders that can present as, and commonly co-occur, with ASD to ensure accurate diagnosis. However, regardless of the presentation of the symptoms, individuals must have the following symptoms specified within the DSM-5 in order to receive a diagnosis of ASD.

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**Deficits in Social Communication and Interactions across Contexts.**According to the DSM-5, individuals with ASD must have deficits with social/emotional reciprocity and communication, nonverbal communication, and building meaningful, lasting relationships with people (American Psychiatric Association, 2013).

*Social/Emotional Reciprocity.* Specifically, individuals with ASD can have difficulty with social/emotional reciprocity, which includes elements of pragmatic communication such as back and forth conversation and social initiation and responses. Social/emotional reciprocity also includes a person's ability to share their interests and emotions with others. Social imitation may be impaired or absent. Notably, the quality of these skills is also evaluated and considered, in addition to the presence or absence of the skills. For example, a skill may be technically present, but the quality of it may be unusual, odd, or inappropriate (American Psychiatric Association, 2013).

*Paraverbal and Nonverbal Communication Difficulties*. Individuals with ASD often have delays and deficits in using and understanding nonverbal social communication, as well as coordinating nonverbal with verbal communication when communicating to others. Examples of nonverbal social communication are eye contact, gestures, and facial expressions. Additionally, individuals with ASD may also have differences in the tone, prosody, and quality of their communicative language. Finally, this symptom category includes an individual's difficulty with demonstrating, communicating, and understanding emotional affect (American Psychiatric Association, 2013).

*Difficulties building, maintaining, and understanding social relationships.* As a result of their core social skills deficits, individuals with ASD often struggle to know how to build lasting, meaningful relationships. There are also times where individuals with ASD also simply prefer to be alone more than a typically developing person. Some researchers think that children with ASD lack a "regional specialization" within the social network of their brain (Johnson, Grossmann, & Kadosh, 2009, p.14). This means when considering the early social skills deficits for children with ASD, it can helpful to think of the individual skills needed to demonstrate the broader skill. For example, parents may report that infants who eventually are diagnosed with ASD may be less 'socially engaged,' which is a broad skill. However, social engagement includes specific skills of eye contact, response to name, attending to faces, and joint attention (Porges, 2003). Individuals with ASD may have delays or deficits any one of these individual skilla reas.

Additionally, children with ASD also often have delays in theory of mind, which can influence their ability to take another's perspective. Theory of mind is someone's ability to understand that people have separate thoughts, opinions, and feelings than themselves. Difficulty with theory of mind along with cognitive inflexibility can influence a young child's ability to engage and access foundational social emotional learning and ultimately build friendships (Mundy & Sigman, 1989). Difficulties seeking out and maintaining friendships can be a major element that impacts the quality of life for individuals with ASD. There are some individuals with ASD who are withdrawn and focused on their internal thoughts and interests versus focusing on others (Buitelaar, 1995; Hepburn & Stone, 2006). However, often there is a desire to have friends, but the variety of skills needed to have those relationships are delayed or not present. Additionally, another aspect of building and maintaining relationships is connecting with supportive people within their life. Individuals with ASD can have difficulty understanding and recognizing when they need to advocate for themselves. Advocacy is another skill that often needs to be directly taught to individuals with ASD (Gallo, Self, & Rausch, 2016).

Finally, children with ASD tend to have limited and/or delayed pretend and collaborative play. Specifically, children with ASD can be literal and concrete in their thinking and pretending something is something else is difficult. Their play can be repetitive, and it can be difficult for them to have flexibility within their play. Many children with ASD are more comfortable engaging in parallel play before they engage in interactive play. Children with ASD who engage in interactive play in early childhood can be 'directors' within their play. Further, their difficulty with shared imagination and engaging in cooperative can impact their ability to connect with peers (American Psychiatric Association, 2013).

**Restricted and repetitive interests**. According to the DSM-5, in order to be diagnosed with ASD, an individual with ASD must have at least two of the four components within the restricted and repetitive interests category. Restricted and repetitive interests is a phrase used to describe a broad range of behaviors that are often uniquely associated with ASD and thus are particularly salient for diagnosis (Watt, Wetherby, Barber, and Morgan, 2008). It can be helpful to think of different levels of complexity of the behaviors. Lower level behaviors are characterized by repetitive motor

movements, speech, use of objects, and self-injury. Higher level restrictive and repetitive patterns of behaviors or interests include insistence on sameness, cognitive inflexibility, and highly restricted interests (Watt, Wetherby, Barber, and Morgan, 2008).

*Stereotyped or repetitive speech and motor movements*. Children with ASD can have delays and/or atypicalities in the development of how they use their verbal communication. These atypicalities can be repetitive or stereotyped and manifest as overly formal ways of communicating, echolalia (e.g. scripted language from others, from a movie, or other form of media without context to explain it), jargon, using rote language, or pronoun reversal. Additionally, individuals with ASD can demonstrate repetitive motor movements such as hand movements (e.g. hand flapping), whole body movements (e.g. rocking), grimacing, skin picking, and other perseverative actions or behaviors. Finally, a third cluster of behaviors under this diagnostic category are related to how individuals use objects. For example, individuals with ASD may demonstrate behaviors such as lining up items/toys, repetitively (compulsively) engaging in different actions like opening/closing doors or turning off/on lights, and nonfunctional play with objects (American Psychiatric Association, 2013).

*Cognitive Inflexibility*. Children with ASD also often have difficulty with cognitive inflexibility in a variety of contexts. Specifically, this can manifest in difficulty adapting to transitions and novelty. Children with ASD often have a 'need for sameness' in all contexts and thrive off routine and consistency. Individuals with cognitive inflexibility can have difficulty with socialization and emotional regulation. For example, children with ASD often find comfort in rules and once they learn a particular

rule they can become extremely upset if they are broken. Inflexibility can also make it difficult for individuals with ASD to engage in play themes, rules, and activities that are outside of what they have learned or what is expected. Finally, rigid, concrete thinking can get in the way of individuals understanding nonliteral speech and humor (American Psychiatric Association, 2013).

*Highly focused interests*. Individuals with ASD commonly have highly specific and intense interests. The intensity of the interest can be described as obsessive or compulsive and they may be overly attached to objects or concepts. Their preoccupations can be repetitive and perfectionistic (American Psychiatric Association, 2013).

*Sensory Differences*. Individuals with ASD often experience hyper and hyposensitivity to sensory input. This can manifest as a high pain tolerance, preoccupation with certain sensory stimuli (e.g. touch, visual, movement), odd or atypical responses to sensory input, and unusual exploration of objects (American Psychiatric Association, 2013).

**ICD-10 Definition of ASD.** The *International Classification of Disease, Tenth Edition (ICD-10)* contains diagnostic codes and was created by the World Health Organization (WHO) for the purposes of documenting the prevalence and variety of disease rates internationally. The ICD is used as a standardized method to conceptualize health information within a variety of healthcare settings that identify comprise national morbidity and mortality statistics by WHO member states. Different governments have developed clinical modifications, but the WHO does not oversee the use of these modifications. Individual organizations within countries who develop the modifications of the manual monitor the use of the modifications. For example, the ICD-10- CM (clinical modification) was developed for the use of the U.S. government by the National Center for Health Statistics (NCHS), Center for Disease Control and Prevention (Center for Disease Control, 2018).

Autism Spectrum Disorder as defined by the ICD-10 aligns more closely to the DSM-IV-TR in the sense that instead of one broad spectrum disorder, it divides Autism Spectrum Disorder into more specific conditions including Childhood Autism, Asperger Syndrome, Atypical Autism, Other pervasive developmental disorders, and Pervasive developmental disorders, unspecified. Other related disorders include Rett's Syndrome, and Childhood Disintegrative Disorder (Autism Asperger's Advocacy Australia, October 2018). Notably, the ICD-10 has not updated terms such as "mental retardation" within its' text. Specifically, similar to the DSM-V and educational identification criteria for ASD, within the ICD-10 definition, Childhood Autism includes impairments of social interaction and abnormalities within their communication abilities (Interactive Autism Network, 2018). The ICD-10 also notes restrictive, repetitive and stereotyped behaviors or interests as part of the diagnostic criteria. Further, for a diagnosis the ICD-10 requires that there is atypicality or delay before the age of three in expressive or receptive social communication, reciprocal social interaction and building attachment, and/or symbolic or functional play (Interactive Autism Network, 2018). Finally, the ICD-10 diagnostic criteria includes several developmental and mental diagnoses to rule out to be sure the symptoms being observed are truly consistent with ASD.

DC: 0-5 Diagnostic Classification of Mental Health and Developmental Disorders of Infancy and Early Childhood. ZERO to THREE is an organization dedicated to research and the distribution of information and resources to promote the health and well-being of infants and toddlers. ZERO to THREE published a unique diagnostic manual in 1994 called *DC:0-3 Diagnostic Classification of Mental Health and Developmental Disorders of Infancy and Early Childhood*, which is the first diagnostic system to use a developmental lens when diagnosing mental health and developmental disorders in early childhood (Weston et al., 2003; ZERO to THREE, 2019). In 2016, an updated manual was published which included current research and a wider age-range. The manual is titled *Diagnostic Classification of Mental health* and *Developmental Disorders of Infancy and Early Childhood: Zero to Five* (DC:0-5) (Klaehn, 2018). These manuals were developed to supplement the information within the DSM-5 (Skovgaard, Houmann, Christiansen, & Andreasen, 2005).

Educational Identification of ASD. Autism Spectrum Disorder (ASD) was added to federal special education law in 1990 and the definition has not changed since (Pennington, Cullinan, & Southern, 2014). The Individuals with Disabilities Education Act of 2004 (IDEA, 2004) defines Autism Spectrum Disorder as a "developmental disability significantly affecting verbal and nonverbal communication and social interaction, generally evident before the age of three that adversely affects a child's educational performance," (Knowledge Inc., 2009). The definition goes on to include repetitive behaviors, atypical responses to sensory stimuli, and a need for sameness. The definition is notably more general than the DSM-5 diagnostic criteria, and the DSM-5 criteria is being incorporated within the educational identification process gradually. As expected with the prevalence of the diagnosis of ASD increasing, the prevalence of the educational identification of ASD has increased as well (Pennington, Cullinan, & Southern, 2014).

*Diagnosis versus identification.* When working with families and considering available resources and intervention plans, it is important to understand the distinction between the medical diagnosis and educational definition of Autism Spectrum Definition (ASD). A medical diagnosis of ASD is given by medical specialists with specialized training in ASD diagnosis (Noland & Gabriels, 2004). Best practice is that a diagnosis is given by a multidisciplinary team of professionals who use a body of evidence to determine symptoms and impact. The multidisciplinary team often includes a psychologist, speech pathologist, and occupational therapist. Other members of the team can include also a medical doctor, psychology intern, and social worker. A medical diagnosis is recognized by insurance companies, and often gives families access to resources (e.g. different therapies and interventions) (Barton et al., 2016; Jensen & Spannagel, 2011). Further, depending on the state, the educational identification of ASD uses the Individuals with Disabilities Education Act, 2004 (IDEA, 2004) instead of the DSM-5 criteria to determine if a child qualifies for special education services within a school setting. One survey indicated that over half of states surveyed used their own definition in place of the federally defined criteria (Barton et al., 2016). Further, a critical component for a child qualifying for an educational identification of a disability, including ASD, is that it has significant educational impact of the disability. If a child

has a medical diagnosis but it is not adversely affecting the child's ability to access the general education curriculum, they would likely not qualify for special education services. However, it is likely for an individual diagnosed with ASD to qualify for an educational identification of ASD (Noland & Gabriels, 2004).

*Regional differences.* It is not mandatory that state educational agencies align to the IDEA 2004 federal definition; however, they must adopt enough of the criteria in order to receive financial support from the federal government (Pennington, Cullinan, & Southern, 2014). Thus, there are significant differences in the definition of ASD used across states to determine educational identification (Pennington, Cullinan, & Southern, 2014). Further, the federal IDEA criteria provide overall evaluation guidelines that include testing in most main areas of functioning that may impact a child's performance within an academic setting including cognitive, academic, communication, motor, and social/emotional. However, each state has leeway in how they follow these criteria which leads to a wide variety of evaluation procedures across states (Pennington, Cullinan, & Southern, 2014). The variability in definitions and evaluation processes can contribute to differences in the reported prevalence of ASD state by state.

Pennington and colleagues (2014) conducted a study that examined the similarity and differences between ASD eligibility criteria per state. They found that the majority of states require impairment in the areas of communication and social interaction that appear before the age of three years of age for children to qualify with an identification of ASD. Some states go into detail regarding the nature of the social and communication difficulties required, while others are more general. Only two states include the specific term 'restricted, repetitive, and stereotyped behaviors and interests' within their definitional criteria. However, all states contain specific elements that fall within the broad category of repetitive and restricted interests such as difficulty with transitions, compulsive or ritualistic behavior, and highly focused interests and stereotyped behaviors (Pennington, Cullinan & Southern, 2014). Further, the study found that it was common for state definitions to contain a component regarding abnormal responses to sensory stimuli (44/50 states). Additionally, most states included exclusionary criteria that included ruling out serious emotional disability (SED). The state criteria often do not account for comorbidity of disorders making it difficult when children present with a complex set of symptoms. Areas of growth and improvement noted were more specific definitions, better, more refined assessment procedures, and improved training for school staff (Pennington, Cullinan, & Southern, 2014).

### Factors that Influence Outcomes and Presentation of the Phenotype of ASD

When conceptualizing outcomes and functioning of individuals with ASD, it is important to consider contextual factors that may influence presentation of symptoms, response to intervention, and outcomes overall. Theories that have an ecological orientation, such as bioecological and developmental models, can be helpful to reference when considering the complex relationships that influence outcomes. Bioecological and developmental models incorporate the concept that individuals develop within a variety of contexts, with interactions of a variety of factors (Silver, Measelle, Armstrong, & Essex, 2005). It follows that the manifestation of disorders and the impact of symptoms evolve over time and in context. They do not emerge suddenly or in isolation (Zahn-Waxler, Klimes-Dougan, & Slattery, 2000).

**Individual factors**. Individual child factors can be considered biological characteristics inherent within an individual. Importantly, symptoms and outcomes can rarely be explained by single individual factors. However, it is critical to understand individual factors so that we can understand them in context.

*Emotional Dysregulation.* Individuals with ASD can demonstrate limited expression, understanding, and regulation of emotions. Additionally, given their intense social skill deficits, tendency for overstimulation, and potential cognitive deficits individuals with ASD often experience social anxiety, and ultimately significant challenges with emotional and self-regulation (Mazefsky et al, 2013; Totsika, Hastings, Emerson, Berridge, & Lancaster, 2015).

*Executive Functioning Difficulties*. One of the reasons for delays in social and emotional milestones with children diagnosed with ASD is that they often have difficulty with executive functioning. Executive functioning is a broad term use to describe functions largely directed by the frontal lobe of the brain such as attention, shifting between tasks, emotional regulation, organization, initiation, and self-monitoring. Neurologically, individuals with ASD do not attend to the same stimuli (e.g. faces) as children who are typically developing, and thus, they do not have the same ways of learning how to cope with emotions and learning social cues as individuals who are typically developing (Mundy & Neal, 2001; Stichter et al., 2010). Some researchers argue that the executive functioning difficulties among children with ASD are directly

related to their difficulty with early social learning (Mundy & Neal, 2001; Stichter et al., 2010). For example, if you are not engaging in joint attention or attending to human faces as much as typically developing peers, you are much less likely to learn appropriate verbal and nonverbal behavior and what responses those behaviors get you. These difficulties can also impact how the environment and people within your environment interact with you (e.g. parent response and style) (Aiken, 2007; Mundy & Neal, 2001).

Additionally, individuals with ASD have inherent difficulty with joint attention (JA). Rutherford and colleagues (2007) define JA is the "triadic coordination of attention between the child, another person, and an object or event," (p.1026). They further argue that JA is a valid measure individual's understanding of others' mental state. It generally develops during play, as an infant develops coordination between their gaze, an object, and an adult. The goal of joint attention is to share an object or experience with another person (Kasari, Sigman, Mundy, and Yirmiya, 1990). JA is thought to be an early form of cultural learning and a precursor of deeper understanding of the thoughts and feelings of others. There are several common aspects of JA that can have later implications on behavior and socials skills. For example, Sheinkopf and colleagues (2004) define initiation of joint attention as behaviors that are demonstrated for the purpose of "social sharing," (p.274). Response to joint attention can be defined as an individual's ability to follow another person's gaze or point. Greater initiation and response to joint attention is generally associated with increased language skills (Casenhiser, Shanker, & Stieben, 2013).

*Learning, Language, and Adaptive Skills Delays or Differences*. In addition to their other delays, it is not uncommon for individuals with ASD to have comorbid learning differences and language delays (Sharma, Gonda, & Tarazi, 2018). Individuals with ASD also often have adaptive skills deficits, even when their cognitive abilities are average or above average (Kalbfleisch & Loughan, 2012; Reis, Baum, & Burke, 2014).

*Cognitive ability.* Intellectual or cognitive abilities are one of the most important contributing factors to the heterogeneity with which ASD presents (Postorino et al., 2016). Generally, cognitive abilities or intellectual functioning are terms that refer to an individual's thinking and reasoning abilities, including verbal, nonverbal, and spatial reasoning and working memory and processing speed. The measurement of cognitive abilities can be controversial and exploring the nuances within the measurement of this construct is beyond the scope of this literature review. However, it is important to understand that the existing measurements of cognitive ability have limitations, particularly for special populations like those with ASD (Postorino et al., 2016). Specifically, standardized measurement can contain bias and may not capture the range of an individuals' unique abilities. Further, like any assessment, it is critical to not reduce a person's abilities to scores and to consider their abilities within context with a body of evidence (Postorino et al., 2016).

Intellectual disability (ID), a term used to refer to what was formally known as mental retardation, is defined by having a Full Scale IQ and overall adaptive skills of a standard score of 70 or below (Postorino et al., 2016; Schalock et al., 2010). In statistics, standard scores are scores that have a common scale, a mean of 100, and a standard deviation of 15 (Postorino et al., 2016). This means that a score of 70 would be two standard deviations below what is consider "Average," or typical for a general population. Thus, intellectual disability is a condition where individuals' cognitive and adaptive abilities are measured to be at least two standard deviations below Average with significant functional impairment (Postorino et al., 2016). The DSM-5 categorizes levels of severity within the diagnosis of intellectual disability based of the individual's adaptive abilities (Postorino et al., 2016). There are the times when the etiology is known (e.g. from a known acquired or traumatic brain injury or congenital condition) and other times the etiology is unknown. There is a broad literature regarding how intellectual disability correlates to an increase in both externalizing and internalizing problem behavior in a variety of populations (Hemmings, Tsakanikos, Underwood, Holt, & Bouras, 2008; Luckasson, 2016).

In the case of ASD, intellectual disability is more common (Matson & Shoemaker, 2009). There are some estimates that the prevalence of intellectual disability is as high as 84% in individuals with ASD with varying degrees of severity (Fombonne, 2003; Postorino et al., 2016; Solomon, Miller, Taylor, Hinshaw, & Carter, 2012). An individual's cognitive ability determines their level of functioning within the ASD diagnosis. If a person has an intellectual disability, it often makes the impact of the disorder more profound. Individuals with ID with ASD are considered to have low-functioning autism (Solomon, Miller, Taylor, Hinshaw, & Carter, 2012).

It is important to gain a thorough and accurate sense of someone's cognitive strengths and weaknesses during the diagnostic process. In general, individuals with

ASD who are no identified with ID, have cognitive and learning differences and can often demonstrate a wide degree of variability within their skills. Individuals with ASD who have higher cognitive abilities tend to have more positive future outcomes (Levy & Perry, 2011). Specifically, individuals with a higher IQ tend to have a higher degree of self-care, communication skills, and ultimately are better able to access education and employment (Levy & Perry, 2011). Notably, when individuals have cognitive abilities that are two standard deviations above average and have ASD, sometimes referred to as twice exceptional, it can present its' own challenges. For example, while their thinking and reasoning abilities may be highly developed, other parts of their development may not be, and in fact they can face extreme challenges in certain areas that coexist with their strengths (Kalbfleisch & Loughan, 2012; Reis, Baum, & Burke, 2014). This discrepancy between their strengths and deficits can create secondary effects, like frustration, that can lead to externalizing and internalizing problems over time (Reis, Baum, & Burke, 2014)

*Gender.* There are many ways to define the complex construct of gender. On a broader, conceptual level gender is defined as the "meanings that individuals and societies ascribe to males and females," (Wood & Eagly, 2010, p.630). There are some researchers who have closely examined gender and how it relates to the presentation of behavior problems. Importantly, researchers who have looked at this concept related to behavioral outcomes have looked at gender in a binary fashion (e.g. male/female) and rely almost exclusively on parent and/or individual report of their biological sex. Moving forward, it will be important to study the concept of gender more inclusively (Kreiser & White, 2014).

With this definitional limitation in mind, some of the previous established relationships within in the literature, beginning around the age of four, find that males are more likely to develop externalizing behaviors than girls. Gender considerations also need to be taken into account when considering the presentation of aggression. In general, physical aggression and antisocial behavior occurs more commonly in males who are typically developing than girls (Liu, 2004 ; Silver, Measelle, Armstrong, & Essex, 2005). This pattern remains true with males with ASD (Kreiser & White, 2014). Further, females with ASD are at greater risk for internalizing problems than those who are typically developing as well as males with ASD. This pattern is consistent with other disability groups as well (Solomon, Miller, Taylor, Hinshaw, & Carter, 2012). Notably, there needs to be more research on girls with ASD in terms of phenotypic presentation and comorbidity (Kreiser & White, 2014; Solomon, Miller, Taylor, Hinshaw, & Carter, 2012).

What we know about gender identity and Autism Spectrum Disorder. Gender identity is an important aspect of self-identity, self-conceptualization, and an individual understanding their role within the larger society (Cooper, Smith, & Russell, 2018; George & Stokes, 2017). In a population of children who are developing typically, developmentally, children start to become aware of their gender as early as 18 months and for sure by the age of three. Further, the majority of children have consistent feelings regarding gender by the time they are within their school age years (George & Stokes, 2017; Van Schalkwyk, Klingensmith, & Volkmar, 2015). Importantly, the process of developing gender identity can continue to change until the end of puberty (Cooper, Smith, & Russell, 2018).

There is little to no available information regarding differences within the developmental trajectory of gender identity within a population of children with ASD; however, delays are expected in most cases (Van Schalkwyk, Klingensmith, & Volkmar, 2015). There is a growing body of research indicating that there are higher rates of expressed gender variance within the population of individuals with ASD than within the typically developing population. Females with ASD tend to experience more frequent and significant gender variance (Cooper, Smith, & Russell, 2018). Individuals who express gender variance are at greater risk of experiencing significant social issues such bullying and purposeful isolation (Cooper, Smith, & Russell, 2018). Further, individuals with ASD have inherent difficulty with socialization and face their own increased risk of being bullied. An individual with ASD who *also* demonstrates diversity within their gender presentation may experience compounded social/emotional difficulties (Cooper, Smith, & Russell, 2018).

### **Psychosocial Factors.**

*Maternal Education*. Maternal education is the only variable within the current study that is not an individual child factor. Specifically, there is a wide body of research that has demonstrated a relationship between psychosocial factors such as maternal age and education level that are predictive of negative outcomes in children including behavior problems and developmental disorders (Laucht, Esser, & Schmidt, 1997). There are some studies that indicate that maternal education can be predictive across

generations on various child physical and mental health outcomes (Augustine, Cavanagh, & Crosnoe, 2009; Carneiro, Meghir, & Parey, 2013). Further the literature indicates that mothers with a higher education level generally invest more time and energy on the care, play, management of activities of their children throughout their development (Carneiro, Meghir, & Parey, 2013; Kalil, Ryan, & Corey, 2012). Further, higher maternal education equips individuals to be better able to access resources within their children's childcare, school, and community, often leading to more positive outcomes (Augustine, Cavanagh, & Crosnoe, 2009; Carneiro, Meghir, & Parey, 2013).

### The Construct of Temperament

While many researchers have devoted their careers to studying temperament, there is ongoing debate about the exact definition, model, and methodology of measuring the construct of temperament. Without a common way to operationalize the construct, it can be difficult to reach a consensus regarding what we know and what are areas for further research. Right now, within a research study or body of work, temperament is defined based of the specific lens of the research or the field of study.

In the spirit of using a common language when studying and discussing temperament, Zentner and Bates (2008) integrated information from most of the current, recognized approaches and created a broad definition of temperament comprised of specific inclusionary criteria and assumptions that a person could use when reviewing literature on temperament. Specifically, Zentner and Bates (2008) posit that there is a clear biological basis (e.g. neurochemical and genetic) for temperament. Further, temperament is comprised of characteristics that should be observable and measurable during the first few years of an individual's life. Additionally, temperament characteristics are generally consistent over time and predictive of outcomes. Individual differences in temperament may be observable in the areas of emotional affect, level of activity, attention, and sensitivity to sensory stimuli (Mervielde & De Pauw, 2012).

Basic forms of temperament can be observed in infancy. A child's temperament is shaped by both genetic factors as well as environmental factors; and the unique elements of a child's temperament may manifest themselves differently depending on the environmental context (Henderson & Wachs, 2007). For example, temperament in newborns can manifest as their level of intensity of distress or avoidance of certain stimuli. However, by the age of three months, differences between frustration and approach behaviors can be observably distinguished through body movement, crying, and smiling. As the child continues to develop, between the ages of four and six months, differences in the child's physical approach to stimuli can be observed. Distinct fear, or what some researchers refer to has behavioral inhibition, develops (versus generalized stress) between the ages of seven and ten months (Bridges, 1932; Kagan, 2013; Krieger & Stringaris, 2015). There is conflicting research regarding how stable temperament is over time. For example, there is some literature that indicates that before preschool age, characteristics of temperament may not be stable, but from preschool through childhood and ultimately adulthood temperament characteristics are consistent and measurable (Martin, 1994). However, other researchers indicate that temperament is generally stable across development and situations (Henderson & Wachs, 2007; Kagan, 1994; Rothbart, 2012).

Theoretically, temperament impacts all aspects of behavior, development, and learning. In general, temperament influences how a person reacts and responds to novelty and adversity (Carey, 1998). A child's temperament is shaped by both genetic factors as well as environmental factors; and the unique elements of a child's temperament may manifest themselves differently depending on the environmental context (Henderson & Wachs, 2007).

### **Differentiating Temperament from other Constructs**

The practical application of temperament into prevention and intervention has been a challenge. Part of the challenge is reaching a clear definition of the construct. It is important to have a clear definition of temperament and to ensure that it has been differentiated from other concepts (Fowles & Dindo, 2009).

**Temperament versus Behavior**. A common misunderstanding regarding temperament is the conceptual overlap with behavior. A helpful distinction between temperament and behavior is that temperament represents the "style and form," the nature, or the "how" of the behavior (Krieger & Stringaris, 2015, p.2). For example, if the child is hitting, temperament would represent the energy taken to hit, or if the child desires a particular object, temperament would represent the child's tenacity and motivation to obtain the toy (Krieger & Stringaris, 2015; Muris & Ollendick, 2005; Rettew, 2013). An individual's temperament influences their emotional, motor, and attentional responses across situations, which can contribute to the quality of the behavior, but is not the behavior itself. Temperament, as the *how* of behavior, is also

different from the *why* of the behavior (the function) and the *what* of the behavior, or the talents or perceptions of an individual (Chess, 1990).

**Temperament versus Personality.** Similar to the concept of behavior, there is often confusion about what makes temperament and personality distinct from each other due to the overlap in the constructs. Another reason that there can be confusion is that like many broad concepts, there is not one common, agreed upon definition of personality. Personality is generally defined by the framework chosen by the researcher, however, as a general rule, it can be helpful to think of temperament as being one of the foundational, core components of what later develops into personality characteristics later in life (Janson & Mathiesen, 2008; Rothbart, 2012a). Personality is generally considered a complex, comprehensive study of a person and temperament is more the behavioral style of an individual (Rothbart, 2012). Personality is a broader concept that emerges later in development, versus the core temperament characteristics available early in life that determine a person's reactions and ability to adjust to their environment and experiences (Gartstein, Bridgett, & Low, 2012; Janson & Mathiesen, 2008; Rothbart, 2012a).

The literature review will now shift to a brief review of the historical background that exists on temperament within childhood, different theoretical perspectives on temperament within a pediatric population, what we know about temperament and Autism Spectrum Disorder, and the existing measures regarding temperament.

### **Brief Historical Background of Temperament in Childhood**

In order to understand any construct, it is critical to understand the historical context. There have been many conceptualizations of temperaments by a variety of cultures dating back to approximately 1500 B.C.E and likely originated in Egypt or Mesopotamia (Flaskerud, 2012). Historically, populations have used temperament for anticipatory guidance for many aspects of their life and they have conceptualized it from a deterministic, biological point of view. Specifically, an early conceptualization of temperament was defined by Hippocrates (circa 460-370 B.C.E.), who argued that temperament was connected to four body fluids or humors: blood, yellow bile, black bile and phlegm. The basic idea was that an imbalance of these fluids led to negative outcomes. He incorporated this theory of humourism into his medical theories (Flaskerud, 2012). Subsequently, Galen (circa AD 129- c. 200), another Greek physician, elaborated on Hippocrates' idea and is often credited with the theory of having four humors conceptualization of temperament. He stated that the four humors, sanguine (blood), choleric (yellow bile), melancholic (black bile), and phlegmatic (phlegm), determined an individual's susceptibility to diseases or specific behavioral or emotional tendencies (Flaskerud, 2012). Galen was the first scientist to categorize and study temperament in detail. It was believed that a person who was within the extreme of these typologies could also have a higher risk of certain mental health conditions. Interestingly, while humourism is no longer actively the used, it has been used as a foundation to talk about and explore temperament by many researchers (Flaskerud, 2012). Further, versions of the idea that biologically based characteristics can influence outcomes can be found from the Middle Ages through the 20<sup>th</sup> century (Rothbart, 2012b).

At the time of Thomas, Chess, Birch, Hertzig, & Korn's (1956) foundational New York Longitudinal Study (NYLS) study, the researchers conceptualized ideas that rebelled against the psychoanalytic and environmentalism approaches to child development of the times (Buss & Plomin, 1986). Specifically, the researchers were interested in the interaction of environmental factors with child characteristics. The research followed 129 individuals for thirty years, primarily from educated upper- to middle- class families living in New York, from the age of four to eight months to adulthood, measuring temperament using a parent interview approach. The families were offered behavioral consultation if maladaptive behaviors emerged over time. Noting the limitations of sample demographics, a secondary study was conducted with Puerto Rican families, which yielded results that were consistent with the original study (Buss & Plomin, 1986; Rothbart 2012b).

Within the NYLS study, semi-structured interviews were used with parents and later, with the parents and the research subjects. For the first eighteen months, parents were interviewed every three months, and then at six-month intervals until the age of five. From age five to age nine, interviews with parents were conducted yearly. From adolescence until age 22, the children were interviewed separately from their parents. Interview topics differed overtime. For example, at age three, a detailed structured interview was conducted regarding parenting practices and how the children cope with novel situations (Chess, 1990; Chess, Birch, & Hertzig, 1960; Mervielde & De Pauw, 2012). In 1963, Thomas and Chess and colleagues began publishing their work and their study set the stage for modern temperament perspectives and future research (Rothbart, 2012a). Findings suggested that temperament was significantly related to later outcomes which will be discussed in more detail later in the document.

### **Theoretical Perspectives Regarding Temperament in Childhood**

There are several perspectives on temperament that are widely accepted and continue to be referenced within the literature. It should be noted that these perspectives are not necessarily in contradiction with one another. They have many overlapping concepts and simply provide different frameworks from which to conceptualize temperament (Calkins, 2005). Specifically, there are researchers who adopt the personality tradition, which conceptualizes temperament from the central assumption that temperament forms the foundation of personality. The individual differences in arousal traditions view temperament as influencing differences in arousal, which in turn influence a person's development and reaction to stimuli. A third broad prospective on temperament can be described as temperament as normal variations in behavioral style. An individual's behavioral style describes how an individual interacts with their external and internal environment (Carey, 1998). The current study is based on this third theory of temperament.

# **Personality Tradition: The Emotionality, Activity, and Sociability (EAS) Approach.** Buss and Plomin (1986) used the work of Diamond (1957) on primates to inspire the temperament criteria within their work with children. Specifically, they argue that temperamental characteristics are inherited, should be present in the first two years of

life, and they should have continuity (e.g. be predictive of later personality traits) (Zentner & Bates, 2008). Further, the three foundational temperament traits that Buss and Plomin (1986) suggested for children are emotionality, activity, and sociability. Specifically, emotionality, is defined as being associated with level of distress or reactivity, activity can be defined a child's level of energy and stamina, and sociability can be defined as an individual's ability to initiate and respond to others appropriately (Zentner & Bates, 2008). Using the emotionality, activity, and sociability (EAS) approach as their framework, Buss and Plomin developed two temperament measures: Colorado Child Temperament Inventory and the Emotionality, Activity, and Sociability Survey for Children (Buss & Plomin, 1984; Krieger & Stringaris, 2015; Zentner & Bates, 2008). In conclusion, the main components of Buss and Plomin's temperament model are that they believed temperament should meet set pre-defined criteria, that it is comprised of three unique elements (emotionality, activity, and sociability), and that it is not easily distinguished from the concept of personality.

### Individual Differences in Arousal: Behavioral Inhibition, Emotional Regulation, or Reactive/Self-Regulation.

*The Behavioral Inhibition Approach.* Kagan (1994) defines temperament as a genetically and physiologically based state that is present as early as three months old and it is generally stable across development (Henderson & Wachs, 2007). He also argues that temperament can influence longer term child outcomes. Kagan categorized temperament as either inhibited (e.g. tentative and cautious) or uninhibited (e.g. spontaneous and outgoing). Kagan and his research team studied behavioral inhibition,

as evidenced by physiological symptoms and observable behavior, when faced with a familiar versus unfamiliar stimuli in infancy and other points of development. (Kagan, 1994; Kagan, Reznick, & Snidman, 1987). The terms 'highly reactive' and 'low reactivity' are used to refer to an infant's motor activity and physiological responses when they are exposed to an unfamiliar situation. Specifically, infants who are highly reactive tend to become inhibited later in life and may be vulnerable to being fearful within unfamiliar events. Other negative outcomes of high reactivity in middle childhood include anxiety, sadness, and somatic symptoms. Low reactivity in infancy can lead to uninhibited behavior within the second year of life (Kagan, 1994; Zentner & Bates, 2008). There are some data to suggest that these temperament responses to novel situations may be consistent later in infancy through at least early adolescence (Kagan, 1994; Kagan, Reznick, & Snidman, 1987). Finally, unique to Kagan's view of temperament is that his ideas of highly reactive or low reactiveness and degrees behavioral inhibition are discrete, biologically based categories (Mervielde & De Pauw, 2012). Kagan also conducted neurological research on brain's reaction to stress within the context of his temperament model (Mervielde & De Pauw, 2012). In terms of measurement, Kagan believed that interviews and questionnaires alone did not give a full or accurate picture of a child's temperament, and he believed that laboratory measurements/studies yielded the most accurate information (Mervielde & De Pauw, 2012). Thus, Kagan conceptualized temperament as genetically and neurologically based. Further, he categorized temperament as being either inhibited or uninhibited and described individual's responses to novel events in the context of reactivity. Finally,

Kagan felt that questionnaires did not give a full picture of temperament went used in isolation (Mervielde & De Pauw, 2012).

## The Emotional Regulation or Temperament as Affect Approach. Goldsmith and Campos (1982) define temperament as individual differences within emotional development, including emotional awareness, expression, and regulation. Specifically, Goldsmith and Campos used the term emotionality to refer to the scope of positive and negative emotion. Their research interest was specifically to explore the differences in the expression of the primary emotions (Mervielde & De Pauw, 2012; Zentner & Bates, 2008). The core emotions Goldsmith and Campos considered were Ekman and Friesen's (1971): "anger, sadness, fear, joy/pleasure, disgust, interest, and surprise," (Mervielde & De Pauw, 2012, p.30). Goldsmith and Campos believed that temperamental characteristics could be measured by codable verbal and motoric behaviors as well as facial expressions (Zentner & Bates, 2008). More recently Goldsmith and Campos and other researchers have also studied the genetic underpinnings of temperament. From a biological perspective, there is some research to suggest neurological contributions to how temperament manifests itself. For example, some studies suggest that changes in the central nervous system contribute to the development of temperamental characteristics over time (Henderson & Wachs, 2007; Mervielde & De Pauw, 2012).

Like Kagan, Goldsmith and Campos felt that a combination of sources of information yielded the most accurate picture of temperament. Thus, Goldsmith and Campos, along with Rothbart, created a laboratory measurement called the Laboratory Temperament Assessment Battery (Lab-TAB) as well as an inventory to gather caretaker ratings regarding child temperament called the Toddler Behavior Assessment Questionnaire-Revised, TBAQ- R (Mervielde & De Pauw, 2012; Zentner & Bates, 2008). Research using the Lab-TAB yielded results that indicated that the stability of temperament is greater within a situation versus across situations. Additionally, children's temperament may present itself differently depending on if the situation is novel or familiar (Henderson & Wachs, 2007).

*The Psychobiological or Self-regulation/Reactivity Approach*. Rothbart defines temperament by exploring the relationship between emotions and emotional regulation, similar to other approaches; however, she put a stronger emphasis on attention and neurobiology (Buss and Plomin, 1986). She elaborates on temperament being a behavioral style to include attention, motivation, and emotions (Mervielde & De Pauw, 2012). Rothbart suggests that there are two fundamental neurological systems that are genetically determined, self-regulation and reactivity, which highlight the interaction between cognitive and emotional development (Henderson & Wachs, 2007; Zentner & Bates, 2008).

Specifically, reactivity can be thought of as the degree of an individual's biological or autonomic arousal (Henderson & Wach, 2007). In contrast, Rothbart (2012a) posits that self-regulation can be defined as behaviors individuals use to control their behavior as a result of positive or negative stimuli. It is the ability to be flexible and maintain attention and arousal that fits the context of the environment. Self-regulation is related to central nervous system function and self-regulation strategies largely depend on the child's developmental level and age. For example, as an infant, a caregiver often

relies on ways to distract the infant or re-direct their attention with a toy. As the child's brain becomes more mature, their attention becomes more flexible and self-directed, and ultimately their ability to self-regulate becomes more advanced and under their control. An individual's level of self-regulation is influenced by their temperament and can influence the responses that they elicit from their environment (Rothbart, 2012a). Further, Ruff and Rothbart (1996) suggest that there is a linear relationship between a child's ability to sustain and shift their attention and their ability to regulate their behavior and emotions. Rothbart considers executive functioning to be a key link to many different outcomes (Rothbart, 2012a) . Behavioral processes related to self-regulation include "approach, avoidance, inhibition, and attentional self-regulation," (Zentner & Bates, 2008, p.11).

Additionally, research conducted by Rothbart and her colleagues (2012a) indicated that regardless of age, temperament can be described by three broad categories surgency, negative affectivity, and effortful control (Mervielde & De Pauw, 2012). Negative affectivity can be defined as a tendency to experience negative emotions like be fear, anger-frustration, or experience social distress. The concept of surgency considers an individual's activity level, seeking sensory experiences, and the positive anticipation of experiencing positive emotions (Krieger & Stringaris, 2015; Zentner & Bates, 2008). Effortful control is defined as an individual's ability to control their inhibitions and direct their attention (Zentner & Bates, 2008). Specifically, effortful control can be considered cognitive strategies for regulating emotions and behaviors such as inhibiting behaviors, monitoring self, and choosing coping strategies (Eisenberg, 2012; Rothbart, 2012a; Zentner & Bates, 2008). As children grow, they develop an increased ability to utilize effortful control, to control their emotions, and regulate their behavior. Evidence suggests that higher levels of effortful control correlate to lower incidents of problem behaviors in the future. A child's degree of effortful control can be influenced by environmental factors such as the nature of the relationship they have with their parents. For instance, cold, overly directive parenting can contribute to lower effortful control. (Ruff & Rothbart, 1996). Consequently, Rothbart and her colleagues created two temperament measures from their work: the Children's Behavior Questionnaire (CBQ) and the Early Adolescent Temperament Questionnaire – Revised for older children (Mervielde & De Pauw, 2012).

In summary, Rothbart's conceptual model of temperament includes that temperament is biologically and genetically based, and conceptually includes reactivity and self-regulation. From a measurement perspective, temperament can be categorized into the three categories of surgency, negative activity, and effortful control. Rothbart has done a considerable amount of research looking specifically at the implications and contributions of attention on development and outcomes (Mervielde & De Pauw, 2012).

**Behavioral Styles Approach: A Foundational Study.** As mentioned above, starting in 1950, Thomas and Chess and their colleagues conducted a foundational longitudinal study, the New York Longitudinal Study (NYLS), that focused on individual child factors and how they contribute to individual differences and outcomes. They conceptualized temperament at the behavioral style of the individual (Buss & Plomin, 1986; Mervielde & De Pauw, 2012). The results of the NYLS yielded nine dimensions of temperament: attention/span persistence, distractibility, quality of mood, intensity of reaction, adaptability, approach/withdrawal, rhythmic (regularity), sensory threshold, and adaptability. Notably, while this research was pivotal for the field and influential to current work, the exact dimensions that resulted from the NYLS are rarely used as they were originally conceptualized (Thomas & Chess, 1986). This earlier paradigm of temperament was criticized for being simplistic. Thomas and Chess responded to the criticism by continuing to evolve their view on temperament, eventually including reactivity and regulatory abilities within their paradigm (Henderson & Wachs, 2007; Konstandtareas & Stewert, 2006).

Further, children who were categorized as slow to warm comprised 15% of the NYLS sample and were defined as being withdrawn at first, slower to adapt, and shy when presented with novel situations. The slow to warm group was described as having a somewhat negative mood, but the intensity with which they responded to novel stimuli was less than those within the difficult temperament group (Carey 1998; Thomas & Chess, 1986). Finally, ten percent of the NYLS sample were children characterized as having a difficult temperament. Thomas, Chess, and their colleagues defined difficult temperament as children who had high intensity reactions, low adaptability, arrhythmicity and general negative mood (Thomas & Chess, 1986). Functionally, these children took a longer time to adjust to new routines, were less likely to take risks, their emotional reactions to situations were often intense (Chess, 1990). The results of the NYLS indicated that children with a temperament categorized as difficult or slow to warm were more challenging to parent and were at higher risk of mental health and behavioral

concerns later in life. They also indicated that children who have had difficult temperament have later adjustment problems both within the home and school environment (Bates, Maslin, Frankel, 1985; Chess, 1990; Lerner & Vicary, 1984; McDevitt & Carey, 1977; Nelson, Martin, Hodge, Havill, & Kamphaus, 1999).

Another main concept that emerged from Thomas and Chess's work was one of the first applications of the bioecological model within the field of child development. Goodness of fit is the idea that there is a meaningful interaction between relational factors (e.g. values, opinions, practices, responses) and child behavior/temperament, which can influence a child's presentation of behavior and ultimately their outcomes (Mervielde & De Pauw, 2012). Thomas, Chess and colleagues (1977) advocated for applying the concept of goodness of fit to intervention and prevention by providing anticipatory guidance to parents regarding their parenting style and behavior strategies based on information gathered regarding their child's specific temperament. When there is what is considered a poor fit between a parent and child this can lead to negative outcomes over time (Snyder, 2004). There are parenting and teaching strategies that can be more effective for children based on their temperament. Specifically, children with difficult temperament tend to be more sensitive to different types of parenting when compared to individuals with easy or average temperament (Bradley & Corwyn, 2008).

In summary, the NYLS study was a foundational study conducted by Thomas, Chess and their colleagues in the field of temperament research. They considered that temperament was the biologically based behavioral style of an individual (Carey 1998; Thomas & Chess, 1986). Based off the results of their research, they also argued that temperament could be categorized generally within different typologies, easy, slow to warm, and difficult. Thomas and Chess (1986) felt there was a reciprocal interaction between individual child factors and the environment, and that temperament had predictive value when considering long term outcomes for children. Further, if the child and the environment were not appropriately matched, this could have direct influence on individual outcomes (Thomas & Chess, 1986).

### **Temperament and Autism Spectrum Disorder**

Similar to the population of children who are neurotypical, research indicates that differences in temperament may be correlated to maladaptive behavior for children with ASD (Eaves, Ho, & Eaves, 1994; Hepburn & Stone, 2006; Konstantareas & Homatidis, 1989). There is notably less literature supporting this theory in the population of individuals with ASD than with children who are typically developing. Further, the behavioral phenotype of ASD is broad and may be impacted by temperament characteristics (Hepburn & Stone, 2006). For example, there are some presentations of the ASD behavior phenotype that include anxiousness, fears, and insistence of sameness. There are other manifestations of the phenotype that include more externalizing behaviors and lack of fearfulness. There is not enough existing literature to full differentiate the characteristics of ASD and temperament or explain how they are related (Hepburn & Stone, 2006).

### **Temperament and Outcomes for Children with ASD**

Many of the factors consistent with the behavioral phenotype of Autism Spectrum Disorder (ASD) can be consistent with what can be considered 'difficult' or highly

sensitive temperament within a typically developing population. The consensus within the literature is that children with ASD have a distinct temperament when compared to typically developing children, as well as those with other neurodevelopmental disabilities. For example, by the age of one year, children with ASD can have difficulty with self- regulation (which can cause intense and more frequent reactions and distress), are often more irritable, have increased negative affect, have difficulty with adapting to novel situations, and can have differences with behavioral inhibition (Schwartz et al., 2009). Additionally, difficult temperament leads to various outcomes with ASD. For example, difficult temperament reported within children of ASD has been shown to exacerbate individuals' abilities to engage and respond socially demonstrated less social engagement and decreased social responsiveness (Schwartz et al., 2009). Further, understanding the nuances of different temperament characteristics for children with ASD may help to clarify the different manifestations of the behavioral phenotype of ASD and the wide range of outcomes for children of ASD (Hepburn & Stone, 2006; Schwartz et al., 2009). Meanwhile, other research has found that children with ASD and other developmental delays have a more difficult temperament than children with neurodevelopmental disorders such as fragile X syndrome, Attention Deficit Hyperactivity Disorder (ADHD), and Down syndrome (Schwartz et al., 2009). Specifically, children with ASD can have less positive affect and fewer adaptive strategies as well as a greater difficulty following directions compared to children with Down syndrome (Schwartz et al., 2009).
Further, children with ASD often have higher emotionality, lower effortful control, and higher reactivity and there is some evidence to suggest that children with ASD have a more negative mood than children who are typically developing (Clifford et al., 2013). Specifically, in one study, negative affectivity best predicted academic functioning and interestingly the individuals who demonstrated the most negative affectivity were higher functioning. Children with ASD who have higher symptomology often have lower effortful control (Clifford et al., 2013)

According to Hartley, Sikora, and McCoy (2008) clinically significant maladaptive behavior causes greater distress than the primary symptoms of ASD. Children with ASD often have comorbid internalizing and externalizing behavior. For example, children with ASD may be more aggressive than typically developing children or children with other developmental disabilities. Lower cognitive abilities, adaptive behaviors, and low expressive language ability often predict increased maladaptive behavior (Hartley, Sikora, & McCoy, 2008). Increased withdrawal behavior is often also correlated social skills deficits. There is some research to suggest that the strongest predictor of externalizing behavior was nonverbal cognitive ability, and the strongest aspect that predicted internalizing behavior was adaptive skills (Hartley, Sikora, & McCoy, 2008; Hepburn & Stone, 2006).

However, in one study, most children with ASD fell within the average range in the temperament domains of activity level, rhythmicity, approach/withdrawal, mood, and distractibility (Hepburn & Stone, 2006). Two-thirds of the sample within the study were reported to have low adaptability, one-half of the sample has mild emotional intensity, and one third were reported to be negative in mood, over one half fell in the nonpersistent range, and one-third were reported to be difficult to distract (Hepburn & Stone, 2006).

The wide variety of results, with a variety of theoretical approaches, highlights the need for more research to further clarifies how temperament manifests within children with ASD, how it is different from the characteristics of the ASD diagnosis, and how the relationships between temperament and outcomes compare to the typically developing population.

#### **Measuring Temperament**

Before preschool age, temperament is not stable, but from preschool through childhood and ultimately adulthood temperament characteristics are consistent and measurable (Martin, 1994). Due to the nature of the available measures on temperament, it is particularly important to consider whether the information that is being gathered is for clinical use or for use in research. This will help guide the measures that are chosen. The psychometric properties of a measure is more important when the measure is being used for research purposes. Measures used for clinical purposes can yield valuable information regardless of the psychometric properties as long as there is a trained professional who knows how to interpret and use the information ethically (Rothbart, Ahadi, & Hershey, 1994; Rothbart & Hwang, 2002). Another factor that helps to guide what measure of temperament to use is the theoretical perspective of temperament. As described previously, the framework of temperament chosen contains certain unique assumptions and terminology, which will ultimately influence how the information is analyzed and applied. See Table 1 for a non-exhaustive list of available measures of temperament within early childhood.

There are a variety of ways to measure temperament, and there is an ongoing debate about what type of evaluation yields the most accurate information. The most common way to measure a child's temperament, particularly in early childhood, are parent/teacher questionnaires and/or laboratory-based measures. Specifically, the available questionnaires regarding temperament are multiple choice and based on recent observations of the child's behavior. Laboratory measures include observing behavior either during structured tasks designed to elicit different aspects of temperament or within more naturalistic settings (Goldsmith & Gagne, 2012; Zentner & Bates, 2008). While measuring temperament through individually administered activities directly with the child in a lab or clinical setting can give valuable information, parent/teacher reports of observed behavior and reactions is more economical and convenient to administer over time (Rothbart & Hwang, 2002). Another strength of parent/teacher report is that the results often yield a comprehensive picture of the child's temperament within the home, school, and community settings across contexts whereas a clinical assessment using laboratory procedures is often one snapshot in time. Limitations of parent/teacher reports can be the parents/teachers may represent their child's temperament in an overly positive or negative way depending on the context of the evaluation and their experiences with the child. Parent/teacher report always lacks objectivity, regardless of the bias (Rothbart & Hwang, 2002).

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If possible, it can be helpful to consider a body of evidence and not rely on one measure in isolation (Gartstein, Bridgett, & Low, 2012; Rothbart, 1981; Zentner & Bates, 2008). However, using multiple pieces of information introduces complications when there are discrepancies between the sources (Gartstein, Bridgett, & Low, 2012). As children grow older, they interact with more professionals in a variety of environments. These professionals can also complete measures regarding the child's temperament and they can have less bias as they are completing the questionnaires since they have less emotional investment and attachment in the results.

Temperament assessment has been limited over the years to specific theoretical perspectives rather than looking at other possibilities (Goldsmith & Gagne, 2012). There are several measures that have resulted from the New York Longitudinal Study (NYLS) and while the internal consistency and reliability of these measures have been reported as low, they continue to be used in present day. There are also several measures based off the psychobiological approach created by Rothbart and her colleagues as well (Gartstein, Bridgett, & Low, 2012). A main difference between measures with a conceptual foundation of the NYLS and psychobiological approaches are that the measures inspired by the NYLS are designed to measure clinically salient temperament characteristics, while the psychobiological measures look more closely at traits that form the neurobehavioral roots of temperament (Gartstein, Bridgett, & Low, 2012).

It is important to note that due to the conceptual overlap of temperament with constructs such as behavior and personality, the results can be skewed. For example, if a child has a high degree of externalizing and internalizing behaviors, there can be a negative skew to the results of the temperament measures (Sanson, Prior, & Kyrios, 1990). Internalizing behaviors in particular have a significant overlap with temperament. Internalizing behavior is defined as being within self and thus "problems" may be seen as on the continuum of internalizing behavior versus distinct constructs (Sanson, Prior, & Kyrios, 1990). While there is considerable overlap with externalizing behavior and 'negative' or difficult temperament traits, the relationship between the two is so strong that psychologists often feel more comfortable considering them distinct constructs. In summary, there needs to me more research clarifying how to truly define and measure these interrelated constructs (Sanson, Prior, & Kyrios, 1990). Sanson and colleagues (1990) emphasized that in order to isolate the constructs completely, there would need to be a very narrow definition of temperament and/or behavior, which is also unhelpful.

### Table 1Measures of Temperament in Early Childhood

<u>Name of the</u> <u>Assessment</u>	Age Range	Author(s) and Dates	<u>Temperament Scales</u>	<u>Type of Reporter</u>
Infant Behavior Questionnaire, Revised (IBQ- R)	Ages 3 months to 12 months	Garthstein & Rothbart, 2003	<u>14 subscales:</u> Approach, Vocal Reactivity, High Pleasure, Smile and Laughter, Activity Level, Perceptual Sensitivity, Sadness, Distress to Limitations, Fear, Falling Reactivity, Low Pleasure, Cuddliness, Duration of Orienting, and Soothability	Caregiver
Children's Behavior Questionnaire (CBQ)	Ages 3 to 7	Rothbart et al., 2001	Positive Anticipation, Smiling/Laughter, High Intensity Pleasure, Activity Level, Impulsivity, Shyness, Discomfort, Fear, Anger/Frustration, Sadness, Soothability, Inhibitory Control, Attentional Focusing, Low Intensity Pleasure, and Perceptual Sensitivity <u>Three Broad Dimensions of</u> <u>temperament:</u> Extraversion/Surgency, Negative	Caregiver

			Affectivity, and Effortful Control	
Infant Characteristics Questionnaire- Revised (ICQ- R) Behavioral Style	Ages three to twelve months 3- 8 years old	Bates, 1982; Gartstein & Rothbart, 2003 McDevitt and Carey.	14 Subscales ; activity level, distress to limitations, fear, duration of orienting, smile/laughter, high pleasure, low pleasure, soothability, falling reactivity, cuddliness, perceptional sensitivity, sadness, approach, and vocal reactivity Activity, Rhythmicity, Approach, Adaptability, Intensity, Mood	Caregiver Caregiver or Teacher
Questionnaire (BSQ) Emotionality.		1978 Buss &	Persistence/Attention Span, Distractibility, Sensory Threshold Emotionality, Activity,	Caregiver
Activity Level, Sociability, and Impulsivity (EASI-III)		Plomin, 1975	Sociability, Impulsivity.	Culogitor
Toddler Temperament Scale (TTS)	1 to 3 years old	Fullard, McDevitt and Carey, 1984	Activity, Rhythmicity, Adaptability, Approach to Novelty, Emotional Intensity, Quality of Mood, Sensory Sensitivity, Distractibility, and Persistence	Caregiver
Revised Infant Temperament Questionnaire (RITQ)	4 to 11 months old	Carey and McDevitt, 1978	Activity level, Rhythmicity, Adaptability, Approach to Novelty, Emotional Intensity, Quality of Mood, Sensory Sensitivity, Distractibility, and Persistence.	Caregiver
Toddler Behavior Assessment Questionnaire (TBAQ)	16 to 36 months	Goldsmith, 1996	Activity Level, Perceptual Sensitivity, Inhibitory Control, Soothability, Appropriate attentional allocation, Sadness, Anger, Interest, Object Fear, Social Fear, Pleasure	Caregiver
EAS Temperament Survey for Children	Ages 18 to 50 months	Rowe and Plomin (1977); Boer and Westenberg, 1994	Emotionality, Activity, and Shyness	Caregiver
Early Childhood Behavior Questionnaire (ECBQ)	Ages 18 to 36 months (supplement to TBAQ)	Goldsmith, 1996	Activity Level/Energy, Attentional Focusing, Attentional Shifting, Cuddliness, Discomfort, Fear, Frustration, High-intensity Pleasure, Impulsvity, Inhibitory Control, Low-Intensity Pleasure, Motor Activation, Perceptual Sensivity, Psitive Anticipation, Sadness, Shyness, Sociability, Soothability	Caregiver
Colorado Childhood Temperament Inventory (CCTI)	Ages 1-6 years old (derived from a combination for the	Rowe and Plomin (1977)	Sociability, Emotionality, Activity, Attention span- Persistence, Reaction to food, Soothability	Caregiver

	NYLS and EASI items)			
Temperament Assessment Battery for Children- Revised (TABC- R)	Ages 3 to 7 years old	Martin, 1998	Activity, Adaptability, Approach/Withdrawal, Distractibility, Emotional Intensity, and Persistence	Caregiver
Childhood Temperament Questionnaire (CTQ)	Ages 3 to 7 years old	Thomas and Chess, 1977	Activity, Rhythmicity, Approach, Adaptability, Intensity, Mood, Persistence, Distractibility, and Threshold	Caregiver
The Dimensions of Temperament Survey (DOTS)	Childhood to Adulthood	Lerner, Palermo, Spiro, & Nesselroade, 1982)	Activity, Rhythmicity, Approach, Adaptability, Intensity, Mood, Persistence, Distractibility, and Threshold as well as "difficult" temperament	Caregiver
The Behavioral Inhibition Questionnaire		Bishop, Spence, & McDonald (2003)	Social Novelty, Situational Novelty, and Physical Challenges	Caregiver
Laboratory Temperament Assessment Battery (Lab- TAB)	Pre- locomotor Version-6 months Locomotor Version – 12 months Preschool Version – 3 to 5 years old	Goldsmith and Rothbart Goldsmith and Rothbart Goldsmith and Reilly	Fear, Anger/Frustration, Joy/Pleasure, Interest/Persistence, and Activity Level Fear, Anger/Frustration, Joy/Pleasure, Interest/Persistence, and Activity Level Fear, Distress, Exuberance, Interest/Persistence, Activity Level, Inhibitory Control, and Contentment	Laboratory Administered Assessment

#### The Construct of Maladaptive Behavior

Psychopathology and dysfunction have behavioral, affective, and cognitive components of their presentation. The current study focuses on behavior within the context of the affective and cognitive components that contribute to negative outcomes (Zahn-Waxler, Klimes-Dougan, & Slattery, 2000). Notably, it is important to understand that behavior in and of itself is not positive or negative. It takes a careful assessment using a multiple assessment points to determine whether the behavior is a sign of a disorder, problem, or dysfunction in general (Carey, 1990; Frick, 2004; Olson, Sameroff, Lunkenheimer, & Kerr, 2009). At an early age, usually between the ages of two and three, behavior that is observable and measurable is apparent and the functional impact of the behavior can also be determined (Mesman, Bongers, & Koot, 2001). A critical window of development in which behavior problems solidify is within the transition of preschool to the school age years (Denham et al., 2000). As social and academic demands increase with age, a child's ability to adapt and cope with changes and selfregulate can determine if they develop increased internalizing or externalizing behavior and ultimately whether they develop negative outcomes.

Conceptually, the terms internalizing and externalizing problems are widely used within the developmental psychopathology literature to further categorize and study maladaptive behavior (Zahn-Waxler, Klimes-Dougan, & Slattery, 2000). There is a broad body of research that exists on externalizing and internalizing behavior and how they relate to various outcomes (Mesman, Bongers, & Koot, 2001; Olson, Sameroff, Lunkenheimer, & Kerr, 2009). Like many concepts within the field of development, externalizing and internalizing problems are most easily described separately but in reality, they are interrelated. For example, there is some evidence to suggest that internalizing or externalizing behavior problems early in life are correlated to longer term internalizing and externalizing disorders. Additionally, internalizing and externalizing behavior can often co-occur and ultimately there is significant covariation between the two concepts (Mesman, Bongers, & Koot, 2001; Rhee et al., 2007; Zahn-Waxler, Klimes-Dougan, & Slattery, 2000).

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**Externalizing behavior.** Externalizing behavior is a broad term used to describe behavior that is observable and usually obvious to everyone around the individual such as aggression, disruptive behavior, defiance, antisocial behavior, and overtly oppositional behavior. Traditionally, this behavior is thought of 'acting out' and impacting those around the individual (e.g. peers, caretakers, other family members etc.). When the behaviors are disruptive or impact others in a negative or harmful way they can be considered a problem behavior or a behavior that is maladaptive (Zahn-Waxler, Klimes-Dougan, & Slattery, 2000). In addition to having an impact on others, a key defining component of externalizing behaviors is that they have a negative emotional component for the individual (Achenbach, 1990; Denham et al., 2000). Specifically, part of the development of externalizing and disruptive behavior can be a related to a deficit in development of self-regulation or other social/emotional skills (Olson, Sameroff, Lunkenheimer, & Kerr, 2009). In early childhood, as expressive communication is developing, children can rely on externalizing behavior, like aggression or opposition, to control the environment around them (Bongers, Koot, Ende, & Verhulst, 2004; Denham et. al., 2000; Sanson, Prior, & Kyrios, 1990).

Without intervention, externalizing problems have strong stability over time (Fagot, 1995; Mesman, Bongers, & Koot, 2001). Early, consistent externalizing behavior not only predicts increased externalizing behavior later in development, but it is correlated to later more pervasive negative outcomes as the child progresses in their development. For example, there is some research to indicate that children who demonstrated hostile and aggressive behavior in early childhood are more likely to have this behavior persist within the school age years (Silver, Measelle, Armstrong, & Essex, 2005). The transition to kindergarten can also be difficult, and often children who have pre-existing externalizing behaviors can struggle exponentially during this time (Silver, Measelle, Armstrong, & Essex, 2005). Further, externalizing behavior are can lead to conduct problems in middle childhood and in adolescence and adulthood outcomes such as juvenile delinquency, crime, and violence. Notably, some parent report data that indicates that behavior naturally decreases over time, however, this may be given that parents can habituate to the behavior over time or there may also be more support for the behavior during the school ages (Mesman, Bongers, & Koot 2001; Silver, Measelle, Armstrong, & Essex, 2005). Externalizing behavior often also takes a toll on the family quality of life, which also ultimately impacts the outcomes of the individual child (Denham, et al., 2000).

An important distinction to make when studying externalizing behavior is that it is different from externalizing disorders. The constructs measured within the current study are limited to externalizing behaviors, which are individual, observable characteristics within the child. Externalizing disorders (e.g. Attention Deficit Hyperactivity Disorder, Oppositional Defiant Disorder, and Conduct Disorder) are a combination of characteristics, or symptoms, that comprise a diagnosable psychological disorder as defined the by the Diagnostic Statistical Manual, Fifth Edition (DSM-5). While this is a recognized area of controversy, a key factor for diagnosing an externalizing disorder is whether the symptoms (in many cases these are behaviors) or characteristics negatively impact their functioning in a variety of areas. Internalizing behavior. Internalizing behavior is a broad term used to can be defined as self-punishing behaviors, over-controlled behaviors, or within-self emotions and moods such as guilt, fear, anxiety, phobias, depression/dysphoria, somatic complaints, withdrawal, and sadness (Hinshaw, Han, Erhardt & Huber, 1992; Quay, 1986; Rothbart, 2012a; Sanson, Prior, & Kyrios, 1990; Zahn-Waxler, Klimes-Dougan, & Slattery, 2000). Internalizing behaviors often do not have the same impact on other people in the same way the externalizing behaviors do. They typically have a more of a negative impact on the individual child than those around them (Zahn-Waxler, Klimes-Dougan, & Slattery, 2000). As children get older, their internalizing behavior problems develop and evolve and make them harder to distinguish from individual behaviors (Suárez, Bennett, Goldstein, & Barlow, 2009).

Similar to the distinction made above, it is important to differentiate internalizing behaviors and internalizing disorders. Two main categories of internalizing disorders are Anxiety and Depressive Disorders. These disorders occur when a group of internalizing symptoms or behaviors reach a threshold where their frequency, intensity and duration have negatively impacted functioning within an individual's life (Bongers, Koot, Ende, & Verhulst, 2004; Brozina & Abela, 2006). There is a high degree of co-morbidity and overlap between internalizing disorders and other psychological conditions (Zahn-Waxler, Klimes-Dougan, & Slattery, 2000).

#### Maladaptive Behavior in Children with Autism Spectrum Disorder

In general, children with ASD often have behavior or emotional regulation problems. For example, in one study with a sample of 143 children, parents reports indicated that 97% of the sample demonstrated externalizing problems and mood lability (Mayes, Calhoun, Murray, Ahuja, & Smith, 2011). It should be noted that maladaptive behaviors rarely occur in isolation, particularly with children with ASD (Mayes, Calhoun, Murray, Ahuja, & Smith, 2011). Some researchers suggest that one of the reasons that some children with ASD develop maladaptive behavior later in life and others do not is because of individual differences within temperament (De Pauw, Mervielde, Van Leeuwen, & Clercq, 2011). Similar to temperament, there is some debate in the literature as to whether behavior problems should be conceptually considered separate or if they are a part of the spectrum of constellation of symptoms that comprise ASD (Hartley, Sikora, and McCoy, 2008). Specifically, externalizing problems, such as aggression, are a significant concern for individuals with ASD (Mazurek, Kanne, & Wodka, 2013). There are very few studies that are on a large scale that involve individuals with ASD. It should be noted that, protective factors such as close relationships have the potential to increase or decrease externalizing behavior depending on the child was able to form an maintain that relationship (Silver, Measelle, Armstrong, & Essex, 2005). Further, some estimates to suggest that over 40% of individuals with ASD have internalizing behaviors that include anxiety (Lenroot & Yeung, 2013). More specifically, individuals with ASD have higher incidence of social anxiety as well as overall anxiety/depressive behaviors (Lenroot & Yeung, 2013). Similar to the typically developing population, certain patterns of early differences in temperament in children with ASD have been shown to continue on within development and lead to later depressive and anxiety symptoms in the future (Schwartz et al., 2009).

#### **Measuring Maladaptive Behavior**

One of the most convenient, common ways to gain information regarding and individual's social/emotional and behavioral functioning is through caregiver or teacher report (Silver, Measelle, Armstrong, & Essex, 2005). As discussed within the temperament section, parent/teacher report data always lacks objectivity and can be skewed based on the context of the evaluation and intensity of the observed behaviors, but can give valuable, comprehensive information. Observational and laboratory measures of maladaptive behavior exist but these are less convenient, time intensive, and not practical. Additionally, trained professionals conducting a file review of past behavior and developmental history as well as parent and teacher interview can be valuable in identifying children who are at-risk for more serious behavior problems later in their life (Silver, Measelle, Armstrong, & Essex, 2005). Finally, functional behavior assessments (FBA) can be conducted by a trained professional gathering a body of evidence on the antecedents, behaviors, and consequences through multiple modalities (e.g. observations, interviews, and file reviews) to hypothesize the function of the maladaptive behavioral (s) and create a behavior plan with interventions and strategies targeting the hypothesized function. Progress monitoring data is collected on the maladaptive behavior to determine the effectiveness of the intervention (or treatment) plan and to adjust it based off the effectiveness of the interventions (Scott et al., 2004). FBAs are rarely used in formal research measure but are commonly used in clinical and school-based settings. See Table 2 for available measures of maladaptive behavior within elementary age children.

## Table 2Available Measurement of Maladaptive Behavior in Elementary Age Children

Name of Assessment	Age range	<u>Author(s),</u> <u>Dates</u>	Behavior Scales	Type of Reporter	
Developmental Behavior Checklist (DBC)	Ages 4 to 18 years old	Einfeld & Tonge, 1992, 1995, and 2002	Disruptive, Self-Absorbed, Communication Disturbance, Anxiety, Social Relating, and Antisocial	Parent and Teacher Report	
Child Behavior Checklist (CBCL)	Ages 1.5 through 5 years old	Achenbach & Rescorla, 1999 and 2000	Three Overall Scales: Total Problems, Internalizing, Externalizing Withdrawn, Somatic Complaints, Anxious/Depressed, Social Problems, Thoughts Problems, Attention Problems, Delinquent Behavior, and Aggressive Behavior	Parent and Teacher Report	
Eyberg Child Behavior Inventory (ECBI) and Sutter- Eyberg Studetn Behavior Inventory- Revised (SESBI- R)	Ages 2 to 16 years old	Robinson, Eyberg, & Ross, 1980	Noncompliance, defiance, aggression, and impulsivity – there is an Intensity and Problem Scale	Caregiver	
Nisonger Child Behavior Rating Form (NCBRF)-	Ages 3 to 16 years	Aman, 1996	Positive Social, Problem Behavior	Parent and Teacher	
intellectual and developmental disabilities	old			Parent	
Nisonger Child Behavior Rating form- Typical IQ (NCBRF- TIQ)		Aman, 2008			
Behavior Assessment System Children, Third Edition	Ages 2 to 21 years	Reynolds & Kamphaus, 2015	Externalizing Behaviors, Internalizing Behaviors,	Parent, Teacher	
(BASC-3)	old		and Adaptive Skills – School Problems (Teacher Report Form)	Self	
Sen- Report Form			School Problems, Internalizing Problems, Inattention/Hyperactivity, and Personal Adjustment		
Aberrant Behavior Checklist	Ages 5 to 51 years old	Aman & Singh, 1986	Irritability/ agitation, lethargy/social withdrawal, stereotypic behavior, and hyperactivity/non- compliance	Caregiver	

Vineland Adaptive Behavior Scales, Third Edition (VABS- 3)	Ages birth to 90 years old	Sparrow, Cicchetti, & Saulnier, 2016	Communication, Daily Living Skills, Socialization, Motor Skills, and Maladaptive Scale (Internalizing and Externalizing)	Parent and Teacher Report
Revised Behavior Problem Checklist (BPC)	Ages 5 to 18 years	Quay & Peterson, 1996	Conduct Disorder, Socialized Aggression, Attention Problems/Immaturity, Anxiety/withdrawal, Psychotic behavior, and Motor Tension/Excess	
Conners Rating Scale- Revised	Ages 6 to 12 years old	Conners, 1970		Parent, Teacher, Self
AAMR Adaptive Behavior Scales, Second Edition		Lambert, Nihira, & Leland, 1993		School and Caregiver
Client Development Evaluation Report (CDER)		California Department of Developmental Services, 1980	Motor Competence, Independent Living Skills, Cognitive Competence, and Social Competence (Adaptive Behaviors) / Social Maladaption and Personal Maladaption (Maladaptive Behaviors)	Caregiver
Scales of Independent Behavior, Revised (SIB-R)	Hill, 1996	3 months to over 80 years		

#### **Relationship between Temperament and Maladaptive Behavior**

Temperament has a large influence over emotional development and behavioral reactions to external stimuli in early childhood as well as on an individual's coping style. There is a high degree of variability within a child's response to their environment as well as their response to novel stimuli, and many researchers indicate that a child's temperament can explain some of this variability (Boyce, 2016; Krieger & Stringaris, 2015). Regardless of the differing perspectives on temperament, it has been well-established in the literature that specific early temperamental styles are strongly related to the development of externalizing and internalizing maladaptive behavioral outcomes later in life (Janson & Mathiesen, 2008; Klein, Dyson, Kujawa, & Kotov, 2012). For example, some research indicates that individuals, particularly males, who have highly negative

temperaments combined with a passive emotional regulation approach are more likely to have internalizing and externalizing problems (Blair, Denham, Kochanoff, & Whipple, 2004). Studies that have been completed looking at temperament and outcomes, indicate that when temperament is measured early in life it also has predictive value when considering different stages of development within the child's life (e.g. preschool, childhood, and adolescence) (Janson & Mathiesen, 2008). Thus, temperament has some important implications on behavior development and adjustment within individuals in general. It provides clues to who is at risk and who can benefit from intervention and prevention efforts (Janson & Mathiesen, 2008; Klein, Dyson, Kujawa, & Kotov, 2012).

Additionally, some researchers say that early temperament can be considered the baseline risk for the individual, and subsequently, experiences within the environment can shape how the temperament characteristics manifests later in life (Klein, Dyson, Kujawa, & Kotov, 2012). Thus, understanding an infant's temperament may have implications for later maladaptive behavior when the child is school age (Janson & Mathiesen, 2008). Researchers suggest that this heightened response to the environment makes it so that children with more difficult, or highly sensitive, temperament may need more external supports to learn coping strategies and regulate their emotions (Boyce, 2016: Belsky & Pluess, 2009). They are more easily aroused negatively and thus are less likely to have inherent abilities (without external help) to organize their environment and utilize the resources around them (Bradley & Corwyn, 2008).

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# Specific Temperament Characteristics Associated with the Development of Maladaptive Behavior

"Difficult" or Highly Sensitive Temperament. "Difficult" temperament traits can also lead to global stress which can lead to behavior problems. The concept of difficult temperament was originally defined by Thomas and Chess, as a result of the NYLS, as negative, intense moods, low adaptability, high frequency of withdrawal to novel situations, and lacking consistency in the biological routines or cycles of eating or sleeping (Carey, 1998; Thomas & Chess 1986; Thomas, Chess, & Birch, 1968). There are other researchers who have used temperament measures like the Carey temperament scales to statistically create 'difficultness composites' by combining temperament characteristics like negative mood and intensity (Bates et al., 1991; Denham et al., 2000). It should be noted that there is only a subset of children with highly sensitive temperament who develop negative problem behaviors, which raises research questions regarding what else contributes to the development of negative outcomes later in life (Denham et al., 2000). Children with difficult temperament have a hard time with internal regulation and are more open to direct assistance with coping strategies (Bradley & Corwyn, 2008).

Overall, having characteristics such as a difficulty adapting to change or novelty, intense emotional expression, pervasive negative mood, or biological irregularity, can put children at higher risk for behavior problems from early childhood to throughout the school age years (Guerin, Gottfried, and Thomas, 1997; Hepburn & Stone, 2006). However, it is important to understand this relationship is not linear, nor is it causal. There are other children with temperament characteristics that would be considered difficulty and they do not develop behavior problems (Guerin, Gottfried, and Thomas, 1997).

**Negative Emotionality.** Some researchers indicate that negative emotionality is most strongly correlated to difficult temperament. High negative emotionality predicts high externalizing and internalizing behavior (Bradley & Corwyn, 2008; Krieger & Stringaris, 2015). Negative emotionality may be considered the "active ingredient in difficultness scores," (Nelson, Martin, Hodge, Havill, & Kamphaus, 1999, p. 697). Negative emotionality as a term can be interchangeable with negative affectivity, difficultness, neuroticism, or reactivity. Traits such as negative mood and high intensity are also likely to contribute to maladaptive behavior because they leads to other things like a 'poor fit' with caregivers or the environment around them (Nelson, Martin, Hodge, Havill, & Kamphaus, 1999). Negative mood and emotionality are often associated with externalizing behavior (Bradley & Corwyn, 2008). Additionally, there is some research to suggest chronic irritability (negative mood) having a relation to depression (Mayes, Calhoun, Murray, Ahuja, & Smith, 2011).

Negative emotions become dysfunctional when there is a high intensity of emotion, duration of emotions, and when emotions are not appropriate to the situation. Individuals with a negative emotional style tend to have reactions to stimuli and environmental factors that are disproportionate (Nelson, Martin, Hodge, Havill & Kamphaus, 1999). These reactions are often externalizing behaviors such as visible anger, defiance, and aggression (Bradley & Corwyn, 2008). Further, individuals with negative emotionality have a more difficult time learning healthy coping strategies then individuals with a less reactive temperament (Bradley & Corwyn, 2008; Krieger & Stringaris, 2015).

Specifically, irritability, a specific aspect of negative emotionality, has been strongly correlated to the development of a wide array of externalizing and internalizing disorders (e.g. Conduct Disorder, Depression, Anxiety, and Oppositional Defiant Disorder) (Krieger & Stringaris, 2015). According to Krieger and Stringaris (2015), negative emotionality is one of the three main ways that temperament can leads to externalizing behavior.

Multiple researchers have used the temperament characteristic of negative emotionality to explain the overlap between the externalizing and externalizing behavior (Rhee et al., 2007). Specifically, Lahey and Waldman (2003) suggest that when internalizing behavior manifests itself as negative emotionality it is a risk factor for externalizing behavior. However, when internalizing behavior manifests itself as a higher ability to control impulses (or low daring) it functions as more of a protective factor. This hypothesis has been supported be several studies (Rhee et al., 2007).

Low Adaptability. Traits like low adaptability are likely to contribute to maladaptive behavior because it leads to other things like a 'poor fit' with caregivers or the environment around them (Thomas & Chess, 1986). Bates (1989) found that low adaptability or negative reactions to novelty early in life predicted internalizing behaviors to a greater degree than externalizing problems.

Effortful Control. Temperament characteristics can lead to externalizing problems when a child has executive functioning difficulties such as low effortful control. Low effortful control is significantly related to many ADHD related behaviors (Rothbart, 2012a). While important considerations, executive functioning and effortful control were not measured within the current study as they are part of Rothbart's (2012) conceptualization of temperament.

**Behavioral Inhibition (related to Approach/Withdrawal).** Behavioral inhibition or fear is a broad sub-dimension of negative emotionality and includes inhibition in response to novel people and situations, avoidance of risks, and/or anxiety about performing in front of others or in situations that involve separation (Kagan, 1994; Zentner & Bates, 2008). In infancy, behavioral inhibition manifests as the degree of tenseness, crying, and motor activity that presents as a response to novel stimuli. Zentner & Bates (2008) report that Kagan posits that a main feature of inhibition is that it is an "intolerance of uncertainty," (p.17), which has been supported by recent neurological research. Notably, behavioral inhibition is different than inhibitory control. Specifically, behavioral inhibition can be considered reactive and fairly automatic distress in new situations. Inhibitory control is a use of executive functioning to regulate or delay gratification. There is literature to support that children who are behaviorally inhibited are more likely to develop anxiety disorders in the future (Suárez, Bennett, Goldstein, & Barlow, 2009).

Temperament characteristics that increase the likelihood of the development of anxiety disorders are withdrawn and inhibited behaviors (Rapee, 2014). Related

temperament characteristics include shyness, fearfulness, and withdrawal. It has been well established within the literature that behavior inhibition, withdrawal and inhibition, in response to social situations, versus open and exploratory behaviors, are correlated to later anxiety (Poole, Jetha, & Schmidt, 2017). There are neuropsychological models of anxiety that also focus on approach and withdrawal behaviors with 'fight-or-flight' in mind (Poole, Jetha, & Schmidt, 2017). Some studies indicate that behavioral inhibition is also correlated to depression (Brozina & Abela, 2006).

**Fear**. There is a fear response that falls within normal limits, and when there is an excessive level of fear or not enough fear, that can be associated with psychopathology (Derryberry & Rothbart, 1997). Excessive fearful behavior is often associated with internalizing behavior or problems (Dewberry & Rothbart, 1997). Notably, fear often loads as negative affectivity or negative emotionality. Other associated terms and concepts are approach behaviors or difficulty with regulation.

#### **Chapter Three: Method**

#### **Study Design**

The proposed study is an *ex post facto* (after the fact), nonexperimental design that involves analyses of archival data collected from previous trials of a large-scale longitudinal study conducted by researchers at JFK Partners, Center of Excellence in Autism and Neurodevelopmental Disabilities in collaboration with the University of Colorado Anschutz Medical School. The original study was entitled: *Longitudinal Study of the Developing Phenotype of Autism*, with principal investigator Dr. Susan Hepburn. Dr. Hepburn also consulted as an expert advisor on the current study.

**Background of the Original Longitudinal study.** Previous to Dr. Hepburn, Dr. Rogers and Pennington were the principal investigators who initiated the longitudinal study in 1996 with the primary goal of exploring the phenotype of autism as comprehensively as possible within a group of young children as part of the Collaborative Programs of Excellence in Autism Network Projects (CPEA), funded by the National Institute of Child Health and Development (NICHD). In 2001, Dr. Rogers transferred to the University of California at Davis, to the M.I.N.D. Institute and mentored Dr. Hepburn. In 2003, the research team received funding to continue and extend the study, with Dr. Hepburn as the principal investigator. Dr. Hepburn continued the study through several more trials at JFK Partners, Center for Excellence in Autism and Neurodevelopmental Disabilities. Notably, the data went through multiple full Institutional Board Reviews through a variety of institutions. The data were collected with integrity and the study design took several precautions to ensure the safety and confidentiality of the vulnerable participants within the sample.

The original sample of children within the study included children from four diagnostic groups: Autism (no known etiology), fragile X syndrome, Down syndrome, and children with developmental disorders with a variety of etiology. There were also typically developing children as participants in the study who were matched on gender, SES, chronological age and mental age. Throughout the scope of the study there were three time points in which data were collected over three developmental periods within the participants' lives: Time 1: Toddlerhood (ages 2-3) ; Time 2 : Preschool (ages 4-5); Time 3: School age (ages 8-11). There were plans to continue the study into adolescence and even adulthood, but the funding sources expired and the resources to sustain a longitudinal study were no longer available. The study originated with collecting data regarding the neuropsychological characteristics of children with autism and related neurodevelopmental conditions. As the study evolved over time and received more funding, an extensive, rigorous testing protocol was designed that looked at child factors, family factors, and environmental factors

#### **Rationale for Current Study**

**Longitudinal Study Design**. Exploring relationships between constructs within the social sciences is often effectively explored through prospective longitudinal study design (Chess & Thomas, 1984). Data collected at different time points demonstrate relationships over time. Longitudinal studies are particularly helpful in exploring how individual child characteristics within early childhood, like temperament, impact outcomes over the course of an individual's life. If the study were cross-sectional or occurring over a more finite period, the long-term, continuous outcomes of the individuals within the study could not be examined (Hoekstra, Happé, Baron-Cohen, Ronald, 2010). Specifically, Happé, (2000) suggests that a longitudinal study design is critical when studying the phenotype of ASD. Burack (2000) elaborates that a longitudinal study design allows an examination of deficits (and skills) that may emerge over time and understanding the sequential nature of the development of the phenotype (or lack thereof). Further, "only a longitudinal study can assess empirically the concurrent and predictive relations amongst various domains of functioning. In addition, a prospective longitudinal design most useful for addressing issues of persistence and precedence of specific deficits within a particular group" (Hepburn, n.d., p.2)

Secondary Data. Importantly, the current study relied on secondary data analysis, which means that the data were already collected prior to the researcher conducting her analyses. Benefits to secondary analysis are that it can be faster and that it minimizes risk to participants from a vulnerable population. Limitations to secondary data analysis include that the theoretical underpinnings and research design decisions are fixed and cannot be modified by the researcher.

#### **Power Analysis**

The power of a statistical test can be defined as "the probability that it's null hypothesis will be rejected given that it is in fact false" (Faul, Erdfelder, Lang, & Buchner, 2007, p.175). In order to determine how many participants were needed to find

statistical significance from the analyses in the current study and to avoid 'false negative' results (a Type II error), power analyses were calculated using the computer program G\*Power 3.1. The program G\*Power 3.1 is a free power analysis program that is stand alone and designed to be used for most of the standard statistical analyses conducted within the social and behavioral fields (Faul, Erdfelder, Buchner, & Lang, 2009; Faul et al., 2007). In social science and educational research, power levels of at least .70 are considered adequate (Stevens, 2007). There are some experts that suggest that in some contexts power levels of .50 are acceptable (Wickens & Keppel, 2004).

A power analysis was conducted for a linear multiple regression, looking at the  $R^2$  deviation from zero to get a sense of the sample size needed for a medium effect size for the overall regression model. A power analysis was also calculated for a linear multiple regression  $R^2$  increase to gain information regarding the sample size needed beyond the sample size yielded from the  $R^2$  deviation from zero analysis in order to measure the effect of temperament above and beyond the control variables within the study. Eight predictors were used within the power analysis to reflect the number of predictors within the study. The results of the first power analysis indicated that in order to achieve statistical power within a moderate or medium effect size a minimum of 91 participants would be required with power of 0.7. Wickens and Keppel (2004) suggest that power of 0.5 can be acceptable and allow the analyses to be robust. When the power analysis was run for a medium effect size with a power of 0.5, a minimum of 65 participants was needed. The power analysis referred to as  $R^2$  increase yielded that in addition to the 91 participants, 70 would be need in order to achieve a moderate or medium effect size a moderate or medium effect size at a

power of 0.7 when looking more closely at the unique influence of temperament within the model. When the same analysis was run at a power of 0.5, 49 additional participants would be needed in addition to the 65 to be able to look more closely at how temperament influences the outcome variables uniquely as compared to the other variables of gender, cognitive functioning, and maternal education. Both at .5 and .7, the current study would be considered underpowered as the available sample was less than the estimated minimum number of participants required.

#### **Decision Rules for Inclusion**

**Rule #1 Diagnostic Criteria**. The raw, de-identified data from the larger sample of children in the longitudinal study which included a children with a wide range of neurodevelopmental diagnoses were used in the study and only children with a clinical diagnosis of Autism Spectrum Disorder as defined by the Diagnostic Statistical Manual, Fourth Edition (DSM-IV) were included within the current sample. Specifically, Autism Spectrum Disorders as defined by the DSM-IV were categorized as Autism, Asperger Syndrome, or Pervasive Developmental Disorder, Not Otherwise Specified (PDD-NOS). The children within the sample needed to meet the diagnostic threshold for ASD on two out of three diagnostic tools: the DSM-IV, the Autism Diagnostic Interview, Revised (ADI-R), and the Autism Diagnostic Observation Schedule, Generic (ADOS-G). The child's diagnosis was confirmed with a body of evidence at each time point within the study. Notably, it was verified that each individual participant included within the study also met the updated diagnostic criteria for Autism Spectrum Disorder (ASD) as defined by the DSM-V (personal communication with Dr. Susan Hepburn, PI, February 2019). Additionally, the children within the study were not diagnosed with any other medical condition nor did they have any hearing or visual impairment.

**Rule #2 Age Criteria**. Secondly, the age criteria for the early childhood time point (Time 1 in the current study), when temperament was measured, included children who at the time of the study were between the chronological ages of two years and five years, eleven months. Temperament is generally considered more stable after two years of life, and therefore the study included not participants under the age of two years (Henderson & Wachs, 2007). Further, the age criteria for elementary school age time point (Time 2 in the current study), when maladaptive behavior was measured, was between the ages of six years and thirteen years.

**Rule #3 Participants at both Time Points**. Each child included in the current study participated at Time 1 and received a full battery of testing during the early childhood period (e.g. developmental testing, family history, diagnostic confirmation, social/emotional functioning, and temperament) and another full battery of testing at school age, Time 3 of the original study, that included parent report of maladaptive behavior on the Developmental Behavior Checklist. Specifically, it was necessary for each participant to have temperament measured between the ages of two years and five years eleven months, as well as maladaptive behavior measured between the ages if six and thirteen years to be included within the study.

**Rule #4 Time Between Assessments**. The time between the temperament measure, Time 1 of the current study, and the behavior measure, Time 2 of the current study, had to be at least 12 months for the child to be included within the sample.

#### **Final Sample Description**

All participants within the current sample met the inclusion criteria mentioned above. Consistent with the national ASD prevalence data, the majority of the sample was Caucasian and male, from moderately to highly educated parents. The sample of children demonstrated a wide range of cognitive and verbal functioning. See Tables 3 and 4 for a summary of the demographic characteristics of the sample.

Table 3.

Variable	Frequency	Percentage	
Race			
African American	4	5.6	
White/Caucasian	63	88.7	
Hispanic	1	1.4	
Missing	3	4.2	
Gender			
Male	57	80.3	
Female	14	19.7	
Maternal Education			
High School	1	1.4	
Some College	11	15.5	
College Graduate	26	36.6	
Graduate	33	46.5	
Diagnosis			
Autism	50	70.4	
PDDNOS	15	21.1	
Aspergers	6	8.5	
Total N	71		

Demographic Characteristics of the Study Sample

Table 4.Mean, Standard Deviation, Skewness, and Kurtosis of Study Variables

Variable	Ν	Mean	SD	Min.	Max.	Skewness	Kurtosis
Time 1: Temperament Measure: Age in Months	71	46.41	10.49	26.00	60.00	35	-1.25
Time 2: Behavior Measure: Age in Months	71	116.55	17.19	77.00	157.00	.39	10
Time Between Assessments in Months	71	70.14	17.57	23.00	112.00	17	.23
Estimated IQ	71	85.70	25.65	35.00	142.00	.07	68
Estimated Verbal IQ	71	79.03	28.91	18.00	144.00	.15	37
Estimated Nonverbal IQ	71	89.07	25.07	36.00	142.00	.07	60

#### **Variable Definitions**

**Dependent (Outcome) Variables**. The outcome variables for the current study were overall maladaptive behavior, represented by the Total Behavior Problem score, externalizing behavior, represented by the Disruptive/Antisocial subscale, and internalizing behavior, Anxiety subscale. Specifically, maladaptive behavior, or behavior problems as they are referred to by the Developmental Behavior Checklist (DBC), is defined as emotion and/or behaviors that supersede a clinical cutoff 58<sup>th</sup> percentile as reported by the parent or caregiver. Additionally, the behavioral observations reported by the child's parent or caregiver must have been observed within six months of completing the assessment (Einfeld, Tonge, & Tonge, 2002). Further, externalizing behavior, as measured by the Disruptive/Antisocial subscale of the DBC within the current study, is defined by parent observation of behaviors like physical aggression, lying, irritable mood, manipulation, and behavior that is considered abusive. Finally, internalizing behavior, as measured by the Anxiety subscale of the DBC within the current study, is defined as parent observation of behaviors such as phobias, fears, crying with ease, feeling distressed when alone, and 'separation anxiety' (Einfeld, Tonge, & Koot, 2002).

Independent Variables. Each temperament construct is defined within the manual of the Carey Temperament Scales (CTS) and these definitions were derived from the results of the New York Longitudinal Study (NYLS). The overall construct, temperament as defined by the CTS is the behavioral style of an individual (Carey & McDevitt, 2000). The individual areas of temperament are given a score between one and four that indicates standard deviations in either direction from the mean. A score of 0.0 would indicate that the child's temperament right at Average for their age. Scores that are positive numbers indicate temperament scores that are more challenging. The higher the numbers, the greater the difficulty and often the more negative impact on the child's life. Scores that are below 0 generally indicate more positive temperament characteristics. According to the test manual, a score of 1.0 can be found in 17 of 100 cases, meaning that +1 and -1 are at the 83<sup>rd</sup> and 17<sup>th</sup> percentiles respectively. Two

standard deviations, or a score of +2 or -2, are considered at the 97<sup>th</sup> and 3<sup>rd</sup> percentiles respectively (Carey & McDevitt, 2000).

Specifically, the temperament subscale of mood is defined as the level of pleasant (or unpleasant) behavior an individual demonstrates in response to certain experiences, situations, and contexts. The construct of intensity is defined as the amount of energy behind individuals' responses "regardless of quality or direction," (Carey & McDevitt, 2000, p.24). Adaptability can be defined as how quickly or gradually a person is able to change their reactions to stimuli in a favorable way. Approach is a construct defined as an individual's initial reaction to novel stimuli (e.g., places, people, situations, things). This item is meant to reflect the degree of inhibition that individual demonstrates towards new stimuli.

**Control Variables.** The available control variables within the archival data were gender, maternal education, and cognitive ability, as represented by estimated IQ. Specifically, in the current study, the construct of gender is defined by parent or guardian report in a binary fashion of whether their child is biologically male or female. Further, the levels of maternal education reported are high school, some college, college graduate, and graduate education. While in the longitudinal study, careful care was taken to capture the participant's cognitive profile, the current study will use estimated IQ as measured by the Wechsler Abbreviated Scale of Intelligence (WASI). An estimate of cognitive ability was made by qualified, doctoral level professionals on each participant based of the WASI constructs of verbal reasoning, nonverbal fluid reasoning, and visuomotor/coordination skills.

#### Measures

The comprehensive test battery of the original longitudinal study includes temperament, diagnostic clarification, developmental, behavior, language, motor, cognitive, and adaptive skills. The descriptions of the measures below are only of the measures relevant to the variables within the study. Specifically, the independent variables are temperament (mood, adaptability, approach, and intensity), cognitive ability, gender, and maternal education. The dependent variables are overall problem behavior, externalizing, and internalizing behavior. Note, in the original study, information regarding the participant's demographics, which included maternal education and gender, was gathered from the parents using a one-page demographic form.

**Temperament: Carey Temperament Scales.** Within the current study, the Toddler Temperament Scales (TTS) or the Behavior Style Questionnaire (BSQ) were used to measure the independent or predictor variables of temperament: mood, intensity, adaptability, and approach. The TTS and BSQ were completed by the parents of the participants to gain more information regarding their early temperament characteristics. The specific form was based on the child. The measures are within a series of Carey Temperament Scales (CTS) designed to measure children's temperament across each developmental level (McDevitt & Carey, 1996).

*Background.* Thomas and Chess and their colleagues conducted a foundational longitudinal study, the New York Longitudinal Study (NYLS), that focused on individual child factors and how they contribute to individual differences and outcomes. They conceptualized temperament at the behavioral style of the individual (Buss & Plomin,

1986; Mervielde & De Pauw, 2012). The results of the NYLS yielded nine dimensions of temperament: attention/span persistence, distractibility, quality of mood, intensity of reaction, adaptability, approach/withdrawal, rhythmic (regularity), sensory threshold, and adaptability. The Carey Temperament Scales assesses temperament within infants and children up to age 12 (Carey, 2001). The CTS series uses Thomas and Chess's nine categories of temperament specifically. In a study conducted by McDevitt and Carey (1977) to standardize the Behavioral Style Questionnaire (BSQ), they found that children who were slow to warm as well as children who were categorized as having a difficult temperament had the following temperament characteristics in common: negative mood, withdrawal, high intensity, and difficulty with adapting to their environment (Carey, 1998). They also replicated the results and that it was more likely that the children with temperament categorized as difficult and slow-to warm would develop increased behavior problems and other negative outcomes later in life (Carey 1998).

*Description of the Measure.* The CTS was chosen for the original longitudinal study due to its strong theoretical backing, the widespread use of the measure in research, the strength of its psychometric properties, and the ease of administration. Items are worded as phrases describing behavior and parents report of how frequently their children exhibit behaviors on a rating scale of one (Almost Never) to six (Almost Always). The measure comprises 100 items and covers nine dimensions of temperament: Activity Level, Rhythmicity, Approach, Adaptability, Intensity, Mood, Persistence, Distractibility, and Threshold of response. The higher the scores on the TTS and BSQ, the more challenging the behavior (Carey & McDevitt, 2000).

Further, per the test manual, overall, summary scores are calculated by "dividing the sum of items on each dimension by the number of ratings available, (Hepburn & Stone, 2006, p.638; McDevitt & Carey, 1996).

*Reliability and Validity*. Per the test manual, validity checks of the results of the questionnaires are done by analyzing missing data, social desirability, and ratings/perception discrepancy (McDevitt & Carey, 2001). Specifically, professionals are alerted when over 20% of the items are missing. When the Average of scores are greater than one in either direction, the social desirability validity score was flagged in order to alert the professional with the clinical interpretation of the measure (McDevitt & Carey, 2001). Notably the authors indicate that if the social desirability scale is elevated, this does not necessarily mean that the rater has bias. It may mean that the child's temperament is extreme and reflects true experiences. Finally, the third validity scale referred to as rating/perception discrepancy is intended to reflect if the rater has an overly positive or negative impression of the child's behavior (McDevitt & Carey, 2001).

Psychometrically, the TTS and BSQ both met reliability and validity criteria for a robust psychological measure that is designed to measure temperament through parent report within three to eight-year-old children. The BSQ was standardized on a sample of 350 children. Studies validating the measure indicated that there was an internal consistency within the dimensions ranging from .47 to .80 (mean of .70) (Hepburn & Stone, 2006; McDevitt & Carey, 1996). Most scores were above .60, with the scales of Rhythmicity and Threshold of Responsiveness being the lowest.

Regarding using the CTS with children with ASD, it should be noted that the norms generally hold, but with some considerations. Specifically, the dimensions of Mood, Rhythmicity, and Threshold of Responsiveness had low internal consistency and thus should be considered with caution when used within a population of children with ASD (Hepburn & Stone, 2006).

#### Maladaptive Behavior: Developmental Behavior Checklist.

Background. The Developmental Behavior Checklist (DBC) was created in 1988 due to an identified need for a standardized measure to assess emotional and behavioral problems in individuals with intellectual disability. At the time, the different measures of functioning for individuals with intellectual disability had various limitations. The authors of the measure wanted to study the prevalence of behavior disorders within this specific population and identify relationships between behavior problems and various outcomes. The intention was to improve intervention planning for individuals with intellectual disabilities and their families (Einfeld & Tonge, 1995). The DBC was designed to reflect behavioral and emotional problems that cannot be explained by a diagnosis of Intellectual Disability (ID). The Developmental Behavior Checklist (DBC) was used within the current study to measure overall problem behavior, externalizing, and internalizing problem behavior in middle childhood. The DBC is a standardized assessment designed to measure behavioral and emotional problems in children and adolescents with a wide range of functioning, including intellectual disability (ID) (Dekker, Nunn, Einfeld, Tonge, & Koot, 2002; Einfeld & Tonge, 1995). Emotional/behavioral assessment of individuals with intellectual disability can be

complicated. Less than 30 years ago, Aman (1991) would not recommend a single measure for the assessment of emotional and behavioral problems because there were no appropriate standardized or field-tested measures available. Aman (1991) mentioned several measures within his article, one of them being the Developmentally Delayed Child Behavior Checklist, which later developed into the DBC. A unique element of the DBC, and one of the main reasons that it was chosen for the current study, is that there are norms created for individuals with a range of cognitive functioning.

Description of the Measure. The measure's 96 items were administered to a sample of 1,093 individuals. Six subscales were obtained from this initial standardization process. These subscales had satisfactory interrater, test-retest agreement, and internal consistency reliability (Einfeld & Tonge, 1995). The items of the measure were developed by examining over 650 case files of children and adolescents who were diagnosed with behavior disorders who had been seen through a developmental assessment service over a period of 12 years. Overall, the norms provided for the measure reflect the prevalence of 96 different behaviors within the community. Each behavior on the checklist given a rating of either zero (not true as far as you know), one (somewhat or sometimes true) and two (very true or often true) (Einfeld & Tonge, 1995). The person completing the form is asked to underline any items that they are particularly concerned about. Then, the examiner can score the DBC on three different levels. First, a measure of overall emotional and behavioral difficulties is the Total Behavior Problem Score synonymous with the Mean Behavior Problem Score. There are also mean and percentiles that can be calculated from the raw scores that indicate the degree of
dysfunction across the five different subscales of disruptive/antisocial, self-absorbed, communication disturbance, anxiety, and social relating. There is a third level that meant to analyze individual items. The DBC can be hand-scored or scored by computer.

*Reliability and Validity.* The measure was originally normed through an epidemiological study of children and adolescents with behavior problems in two Australian States (Dekker, Nunn, Einfeld, Tonge, & Koot, 2002). Dekker and colleagues (2002) reassessed the factor structure of the revised DBC using a large, diverse sample of individuals with intellectual disability (n=1536). Specifically, parent and teacher DBC ratings of Australian and Dutch individuals were combined (ages 3-22 years; mean age 12.1 years). The individuals in the sample displayed a range of functioning, from mild to profound intellectual disability. The results of the study confirmed that the DBC has robust reliability and validity. Specifically, elements such as test-rest reliability, construct validity, and criterion-related validity were all strong (Dekker, Nunn, Einfeld, Tonge, & Koot, 2002).

#### **Cognitive Functioning: The Wechsler Abbreviated Scale of Intelligence**

(WASI). An assessment of cognitive ability was conducted at each time point within the original longitudinal study. At the earlier time points, the Mullen Scales of Early Learning (MSEL) was administered to the participants to evaluate their developmental functioning. At later time points, cognitive tests such as the Differential Abilities Scales, the Leiter International Performance Scale, or the Wechsler Abbreviated Scale of Intelligence (WASI) were attempted to gain a true sense of the individuals cognitive

abilities based of the child's language level and ability to participate within the assessment (personal communication with Dr. Susan Hepburn, PI, February 2019).

*Description of the Measure*. The Wechsler Abbreviated Scale of Intelligence (WASI) is an individually administered assessment designed to measure an individuals' overall thinking and reasoning or cognitive abilities in children ages 6 years to 89 years of age. Specifically, the WASI has four subtests that measure skills in the areas of verbal reasoning, spatial reasoning, and nonverbal reasoning (Stano, 2004). These four subtests yield an overall score referred to as the Full Scale Intelligence Quotient (FSIQ). The WASI is considered a screener and is often administered a full cognitive battery. For research purposes, it can be considered a shortened version of the Wechsler Intelligence Scale for Children, Fourth Edition, with the understanding that it is an estimate, not a comprehensive measure, of a person's overall cognitive abilities (Stano, 2004)

*Reliability and Validity.* The WASI has robust reliability and validity (Stano, 2004). The WASI is considered to have a degree of convergent validity with the WISC-IV that is satisfactory, and while they are not interchangeable, the WASI can be used as a reliable and valid estimate of a child's cognitive ability (Canivez, Konold, Collins, & Wilson, 2009; Philofsky, Fidler & Hepburn 2007).

**Demographic Control Variables: Child Information Form.** Demographic information was collected at each timepoint using the unpublished Child Information Form as designed by Wehner (1996) and colleagues and the University of Colorado. The form was completed by the parent or caregiver before or during each testing session as part of a packet of questionnaires that they completed as their child was being assessed (personal communication with Dr. Susan Hepburn, PI, February 2019).

Inclusion Criteria: Autism Diagnostic Measures. The gold standard in ASD diagnosis was used to confirm the diagnosis of each participant. Currently this standard is that the child is given a comprehensive assessment that includes the Autism Diagnostic Observation Schedule (ADOS) as well as Autism Diagnostic Interview, Revised (ADI-R). Notably, there is an updated version of the ADOS that was created in response to the updated DSM-V diagnostic criteria called the ADOS-2. However, within the current study the ADOS was used as well as the DSM-IV criteria for ASD. Analysis and implication considerations will be discussed in more detail later.

*Autism Diagnostic Observation Schedule (ADOS).* The ADOS is an individually administered, semi-structured assessment of an individual's social, communication, and play skills. A unique part of the ADOS is that it has four modules or versions that are determined by the clinician based on language and developmental level. Depending on the age and level of functioning of the individual, a caregiver is often in the room (Lord, Rutter, DiLavore, & Risi, 2008). The ADOS requires extensive specialized training and a thorough understanding of the characteristics of ASD in order to be administered with validity. The ADOS consists of a series of carefully designed, standardized tasks that are administered to the individual and the individual's behaviors are observed and carefully coded by the administrator. The structured and unstructured tasks are designed to elicit social behavior and communication. The quality and quantity of an individual's skills, depending on the specific skill, are scored. There is technically a cutoff score that the

ADOS yields; however, the authors of the measure are clear that this score should not be used in isolation to determine an ASD. The ADOS is one tool that is to be used within a body of evidence (which includes a thorough developmental history) to determine if an individual meets the diagnostic criteria for ASD and if these symptoms are causing functional impairment within the individual's life (Lord, Rutter, & Le Couteur, 1994; Lord, Rutter, DiLavore, & Risi, 2008).

*Autism Diagnostic Interview Revised (ADI-R).* The ADI-R is a standardized interview designed to be administered to a caregiver in order to get a thorough developmental history with a specific focus on the early characteristics of ASD. The items are administered verbally to the caregiver and the responses are coded by the administrator. Items are organized according to three areas: Communication/Language, Reciprocal Social Skills, and Rigid and Repetitive Behaviors/Interests. Similar to the ADOS, the measure has an algorithm that yields cutoff scores. However, it is discouraged that these cutoff scores be used in isolation within the diagnostic process (Rutter, Le Couteur, & Lord, 2003).

#### **Plan for Data Analysis**

The plan for data analysis was quantitative and comprised of two main statistical processes to answer the research questions and address the hypotheses, Pearson correlation and hierarchical multiple regression.

**Preparation for Analysis**. The first step in the analysis process, was preparing the data for analysis. The steps to prepare for analysis included: addressing any missing data, running descriptive statistics, addressing outliers, and making sure the assumptions

were met for each analysis process. Specifically, normality, linearity, multicollinearity, homogeneity of variances, and homoscedasticity (the distance between the data points and a straight line) were examined to ensure that the analyses could be run (Tabachnick & Fidell, 2013). Establishing the validity and reliability of the Carey temperament measure was considered within the current study, however item data were not available.

**Establish Relationships between the Variables through Correlation.** In order to address research question one regarding establishing relationships between the variables, zero order correlations were used to determine the strength of relationship between the independent and dependent variables (Gliner, Morgan, & Leech, 2009; Schwartz et al., 2009). Specifically, correlations were run to explore parent report of child temperament collected in early childhood and overall, internalizing, and externalizing maladaptive behavior within the elementary school age years. Relationships were also explored between the independent variables of cognitive ability, gender, maternal education and later maladaptive behavior.

#### Hierarchical Multiple Regression to Explore Relationships Between the

**Variables.** In order to answer the second research question regarding what predicts outcomes, hierarchical multiple regression was used to determine if independent variables (e.g., temperament, gender, cognitive ability, maternal education) predicted the dependent variables of overall problem behavior, externalizing, and internalizing problem behaviors (Gliner, Morgan, & Leech, 2009). Hierarchical regression is used when a researcher is looking at the strength of a predictor when compared to other predictors (Petrocelli, 2003). After the assumptions were tested and met, block one of the

hierarchical regression was factors that were controlled for: cognitive ability, gender, and maternal education. The second block of the regression consisted of the individual temperament factors of adaptability, approach, intensity, and mood. The outcome, or dependent, variables were overall problem behavior, externalizing and internalizing behavior. Three separate hierarchical regressions were conducted.

## **Research Questions**

**Question One.** What specific temperament characteristics in early childhood are related to overall, internalizing, and externalizing maladaptive behavior in later childhood in a sample of children with Autism Spectrum Disorder (ASD)?

*Hypothesis One.* There is a statistically significant correlation between the temperament subcategories of: adaptability, intensity, mood, and approach identified in early childhood and overall, internalizing, and externalizing maladaptive behavior later in childhood in a group of children identified with ASD.

**Question Two.** After accounting for maternal education, gender, and cognitive functioning, do the specific temperament characteristics of adaptability, intensity, approach, and mood significantly predict overall, internalizing and externalizing maladaptive behavior later in childhood in a group of children with ASD?

In general, it is hypothesized that early temperament characteristics significantly predict maladaptive behavior later in childhood above and beyond maternal education, gender, and cognitive functioning in children with ASD. Specifically:

*Hypothesis 1.* Controlling for maternal education, gender, and cognitive functioning, overall, internalizing, and externalizing maladaptive behavior in later

childhood in children with ASD is statistically significantly predicted by the temperament characteristic of adaptability.

*Hypothesis 2.* Controlling for maternal education, gender, and cognitive functioning, overall, internalizing, and externalizing maladaptive behavior in later childhood in children with ASD is statistically significantly predicted by the temperament characteristic of intensity.

*Hypothesis 3*. Controlling for maternal education, gender, and cognitive functioning, overall, internalizing, and externalizing maladaptive behavior in later childhood in children with ASD is statistically significantly predicted by the temperament characteristic of mood.

*Hypothesis 4.* Controlling for maternal education, gender, and cognitive functioning, overall, internalizing, and externalizing maladaptive behavior in later childhood in children with ASD is statistically significantly predicted by the temperament characteristic of approach.

*Hypothesis 5.* Controlling for maternal education, gender, and cognitive functioning, overall, internalizing, and externalizing maladaptive behavior later in childhood in children with ASD is statistically significantly predicted by the combination of temperament characteristics of adaptability, intensity, mood, and approach.

Table 5Research Questions and Hypotheses

Research Questions and Hypotheses	Planned analysis technique								
<b>Question 1.</b> What specific temperament characteristics in early childhood are related to overall problem behavior, internalizing, and externalizing maladaptive behavior in middle childhood in a sample of children with Autism Spectrum Disorder?	Establish Relationships using Correlation								
<i>Hypothesis 1.</i> There is a statistically significant correlation between the temperament subcategories of: adaptability, intensity, approach, and mood identified in early childhood and overall, internalizing, and externalizing behavior problems later childhood in a group of children identified with ASD.									
<b>Question 2.</b> After accounting for maternal education, gender, and cognitive functioning, do the temperament characteristics of low adaptability, high intensity, low approach, and negative mood significantly predict overall, internalizing and externalizing problem in middle childhood in a group of children with Autism Spectrum Disorder?	Hierarchical Multiple Regressions								
In general, it is hypothesized that early temperament characteristics significantly predict maladaptive behavior in middle childhood above and beyond the individual factors of maternal education, gender, and cognitive functioning in children with ASD. Specifically:									
<i>Hypothesis 1.</i> The temperament subcategory of adaptability identified in early childhood is a significant predictor of overall, internalizing, and externalizing maladaptive behavior in later childhood in a group of children identified with ASD.									
<i>Hypothesis 2.</i> The temperament subcategory of intensity identified in early childhood is a significant predictor of overall, internalizing, and externalizing behavior problems in later childhood in a group of children identified with ASD.									
<i>Hypothesis 3.</i> The temperament subcategory of approach identified in early childhood is a significant predictor of overall, internalizing, and externalizing maladaptive behavior in later childhood in a group of children identified with ASD.									
<i>Hypothesis 4.</i> The temperament subcategory of mood identified in early childhood is a significant predictor of overall, internalizing, and externalizing behavior problems in later in a group of children identified with ASD.									
<i>Hypothesis 5.</i> Controlling for maternal education, gender, and cognitive functioning, overall, internalizing, and externalizing maladaptive behavior later in childhood in children with ASD is statistically significantly predicted by the combination of temperament characteristics of adaptability, intensity, mood, and approach									
Variables:   Independent (predictor): temperament characteristics- approach, mood, intensity, adaptability   Dependent (outcome): overall maladaptive behavior, externalizing behavior (disruptive/antisocial)   behavior (anxiety)   Control: maternal education, gender, cognitive ability (estimated IQ)	, internalizing								

## **Chapter Four: Results**

# Introduction

To answer the research questions regarding the relationships between temperament variables and maladaptive behavior as well as the specific predictive relationships between the individual temperament variables and the maladaptive behavior variables, all statistical analyses were conducted using the Statistical Package for the Social Sciences version 23 (SPSS 23). The statistical procedures were all two-tailed tests of significance with an alpha level of p < .05. Approval for the study was obtained from the University of Denver's Institutional Board of Review on 2/14/2019 (IRB Approval Number: 731918-1).

## **Preparation for Analysis**

Addressing Missing Data. Ultimately, there were 72 participants who met the inclusion criteria regarding diagnosis, age, participating at both time points needed, and having at least one year between their measure of temperament and maladaptive behavior. In preparation for the analysis process, frequencies were run and analyzed in order to identify and explain any missing data. There were no missing data within any of the independent, dependent, and control variables within the study.

**Outliers.** Outliers are critical to identify, because extreme values can directly influence and skew the results. Specifically, outliers can influence the accuracy of

means, cause an increase in Type I and II errors, and limit the generalizability of the finding (Garson, 2013). Often outliers are eliminated from the sample; however, if the outliers are not extreme and if they are less than 2% of the sample then it is acceptable to leave them in the sample (Cohen, West & Aiken, 2014).

*Univariate Outliers*. In order to identify univariate outliers, histograms and boxplots were created and visually examined for the independent, dependent, and the one continuous control variable (cognitive ability). The data were also visually inspected for extreme values. Upon examination, it was noted that there were a few cases that had outliers in the areas of Anxiety, Adaptability, Intensity, and Mood but none of them were flagged as extreme and thus were kept within the sample.

*Multivariate Outliers*. Multivariate outliers were identified with Mahalanobis distance. Mahalanobis distance reflects how much the vector of case responses differs from the vector of means of the variables (Tabachnick & Fidell, 2013). Tabachnick and Fidell (2013) propose significance at  $\alpha \leq .001$  be used when determining outliers. Using syntax through SPSS to first calculate the chi-square *p* values for Mahalanobis distance and then to flag any critical values, one case was identified as a significant outlier. The multivariate analyses (the multiple regressions) were run both with this case included and without this case in order to ensure the robustness of results. Notably, when the multivariate outlier was removed, the results changed to a degree that the researcher decided to present the analysis results with that case removed. The new sample size was 71.

Assumption of Normality. The assumption of normality was examined across the continuous variables within the study through examining skewness and kurtosis. Skewness reflects how symmetrically the data are distributed (Gliner, Morgan, & Leech, 2009). Values between one and negative one reflects a degree of skewness that meets the assumption of normality. Kurtosis reflects the shape of the data curve. Data that are normally distributed are "neither too peaked nor too flat and its tails are neither too short nor too long" (Gliner, Morgan, & Leech, 2009, p.147). Acceptable values of kurtosis are between three and negative three. Upon examination of the skewness and kurtosis statistics, all values fell within the range that indicated that the assumption of normality was met (see Table 5). Specifically, the temperament variables of mood, adaptability, approach, and adaptability all had a normal distribution. Further, the maladaptive behavior variables of overall behavior problems, disruptive/antisocial behavior, and anxiety were also normally distributed. Additionally, for all the regression models, the Normal P-P Plot yielded a relationship between the variables that were roughly straight line with no significant deviations from normality.

Assumption of Linearity. Correlation and regression assume linear relationships between variables; therefore, for both research questions linearity was established. Scatterplots were used to assess the linear relationships among the variables. Visual inspection indicated no violations of linearity across the individual relationships between the variables (Garson, 2013).

## **Explanation of Scores Used Within the Analyses**

**Temperament**. The scores yielded from administration of the Carey Temperament Scales (CTS) are scaled scores. Specifically, for each of the individual areas of temperament the sum of each of the items reported is divided by the number of available items and this yields a summary score. The summary scores ultimately yield scaled scores either between 1 and 4 or -1 and -4. A score of 0 would indicate temperament that is exactly typical compared to other same-age children. The farther away from 0, either positive or negative, indicate the more challenging the temperament with presumably more negative impact on the child. These scaled scores are used within the analyses (Carey & McDevitt, 2000).

**Maladaptive Behavior.** The Developmental Behavior Checklist (DBC) yields means and percentiles that are calculated from raw scores. The means were not entered in the archival data, and so could not be used in the analyses. The percentiles were hand calculated and entered by the researcher; however, after conversation with the principle investigator, Dr. Susan Hepburn, the raw scores were chosen as a more accurate representation of the degree of maladaptive behavior reported by the caregiver regarding their child (Dr. S. Hepburn, personal communication, 2/18/2019). The greater the number, the more maladaptive behavior is indicated. Importantly, there is an overall Total Behavior Problem score that is a summary score yielded from five different areas of maladaptive behavior examined by the DBC, including the two subscales examined within the study Disruptive/Antisocial and Anxiety. **Control Variables.** Finally, cognitive ability was examined by using the estimated Full Scale IQ (FSIQ) demonstrated by the child on the Wechsler Abbreviated Scales of Intelligence (WASI). The estimated FSIQ is represented by a standard score which is a continuous variable. Maternal education and reported gender of the children within the study are both categorical variables. Specifically, gender was reported in two categories by the caregiver: male or female. Further, maternal education was reported on the demographic form within the original study in four categories: high school, some college, college graduate, or graduate level education. When frequencies were run by the researcher within the analysis preparation, the results yielded only one mother who reported being a high school graduate, which was an outlier. Given the low power of the study, removing one case would lower the power to a greater degree. Thus, new category was created "high school or some college" for the purposes of maintaining power within the multivariate analyses.

#### **Research Question One: Correlation**

Pearson correlations were used to explore the specific relationships between the four temperament variables of adaptability, intensity, mood, and approach and maladaptive behavior (overall, disruptive/antisocial, and anxiety). It was hypothesized that there are statistically significant correlations between the temperament subcategories of adaptability, intensity, mood, and approach identified in early childhood and overall, internalizing, and externalizing maladaptive behavior in later childhood in the available sample of children with ASD. The results of the Pearson correlation analyses are reported below.

First, the relationship between mood and overall maladaptive behavior was positive, with low-moderate strength, and statistically significant (r (69)=.32, p=.01). Similarly, there was a low-moderate positive relationship between the temperament subcategory of intensity and disruptive/antisocial behavior, which was also statistically significant (r (69) = .32, p = .01). There was also a positive, weak correlation between mood and disruptive/antisocial behavior (r (69) = .31, p = .01). Further, there was a positive weak relationship between the temperament subcategory of approach and anxiety that was statistically significant (r (69) = .30, p = .01). Additionally, there was a positive weak association between temperament subcategory of mood and anxiety, and it was statistically significant (r (69) = .26, p = .03). Notably, there were no other statistically significant correlations (See Table 7).

#### Table 6

Descriptive S	Statistics for	<sup>.</sup> Independen	it and Depende	ent Variables
1		1	1	

Variable	Ν	Mean	SD	Min.	Max.	Skewness	Kurtosis
Independent Variables:							
Temperament							
Approach	71	3 59	1.06	1 36	5 64	-0.10	-0.90
Adaptability	71	3 78	0.78	1.90	6.00	0.08	0.37
Intensity	71	3.70	0.70	2.00	5.20	0.00	0.27
Maad	71	2.62	0.70	2.00	5.20	-0.38	0.22
Mood	/1	3.02	0.82	1.00	5.50	-0.30	0.57
Dependent Variables: Maladaptive Behavior							
Total Behavior	71	58.93	23.93	7.00	118.00	-0.16	-0.31
Problem	71	17.17	8.44	1.00	37.00	0.20	-0.60
Disruptive/Antisocial Anxiety	71	7.21	3.43	0.00	15.00	0.71	0.44

## Table 7

# Pearson Correlation Coefficient for Continuous Control, Independent, and Dependent Variables (n=71)

Variable	1	2	3	4	5	6	7	8
1. Approach	1.00							
2. Adaptability	.32**	1.00						
3. Intensity	11	.06	1.00					
4. Mood	.24	.49**	.19	1.00				
5. Total Behavior	01	.07	.16	.32**	1.00			
6. Disrupt/AntiSoc	03	.18	.32**	.31**	.72**	1.00		
7. Anxiety	.30*	.04	.15	.26*	.47**	.30*	1.00	
8. Estimated IQ	.19	.20	.11	.14	20	.01	.26*	1.00

\*p < 0.05 (2-tailed); \*\*p < 0.01 (2-tailed)

## **Research Question Two: Multiple Regression**

In order to answer the second research question regarding the predictive relationships between the temperament and behavioral variables, hierarchical multiple regression was used. There were five hypotheses related to research question two which will be outlined individually as the results are reported. In general, it was hypothesized that early temperament characteristics significantly predict maladaptive behavior later in childhood above and beyond the individual factors of maternal education, gender, and cognitive functioning in children with ASD. The factors of maternal education, gender, and cognitive functioning were controlled for in the regression models.

Assumptions. The assumptions of normality, linearity, homoscedasticity, and mean independence were confirmed. The researcher also found no multicollinearity or significant outliers within the sample (Tabachnick & Fidell, 2013). Specifically, as outlined by Tabachnick and Fidell (2013), normality was examined by graphing the residuals of the individual regression models using histograms and information from the normal curve. The histograms were inspected visually, and it was determined that the residuals for the regression models followed a normal distribution to a sensible degree. Thus, it was determined that the assumption of normality was met.

The assumption of mean independence has multiple factors. First, it is important that any independent variables that may influence the outcome variables are included within the regression model. These variables should be determined by a comprehensive review of the literature. In this study, cognitive ability (estimated IQ), maternal education, and gender were included in the regression model as control variables as the literature indicates a significant relationship between these factors and maladaptive behavior. Further, the constructs need to be measured accurately, and thus the measures chosen need to have robust reliability and validity. The measures of the independent and dependent variables were all chosen for their robust psychometrics. Item data was not available to run specific reliability or validity checks on the specific participants within the sample. Further, the assumptions of homoscedasticity as well as mean independence were also examined through a visual inspection of scatterplots. It was confirmed that the distribution of the residuals was not curvilinear nor was it cone shaped. The data points were generally concentrated in the center and evenly distributed on both sides of the center line. Thus, these assumptions were met (Tabachnick & Fidell, 2013).

Additionally, the Durbin-Watson coefficient d values were analyzed for each regression model to examine further the independence of errors assumption (Garson, 2013). While there is variation in what is considered an acceptable cutoff for the Durbin-Watson statistic, there are some statisticians who indicate that the d value should fall between 1.5 and 2.5 to meet the independence of errors assumption (Garson, 2013). Per this criteria, the independence of errors assumption was met for all 15 regression models. Please see Table 8 for the specific d values.

Table 8

	<b>Regression Models</b>	Durbin-Watson Values (d value)
1.	Approach – Total Behavior	1.91
2.	Adaptability—Total Behavior	1.93
3.	Intensity – Total Behavior	1.96
4.	Mood—Total Behavior	2.01
5.	All Total Behavior	2.00
6.	Approach—Disruptive/Antisocial	1.57
	Behavior	
7.	Adaptability—Disruptive/Antisocial	1.64
	Behavior	
8.	Intensity—Disruptive/Antisocial	1.52
	Behavior	
9.	Mood—Disruptive/Antisocial Behavior	1.73
10.	All Disruptive/Antisocial Behavior	1.62
11.	Approach—Anxiety	1.98
12.	Adaptability Anxiety	1.87
13.	Intensity—Anxiety	1.96
14.	Mood—Anxiety	1.76
15.	All Anxiety	2.03

**Durbin-Watson Values** 

Further, the multicollinearity assumption was examined using the variance inflation factor (VIF) as well as tolerance. Tolerance indicates the degree of variability of the specific independent variable is not explained by the other independent variable within the model. The VIF statistic is the inverse. These numbers are a way to formally check whether the predictors within the study are not too highly correlated. For the assumption to be met, tolerance score needs to be above 0.2 and the VIF score need to be below 10 (O'Brien, 2007). Based on either tolerance or VIF, this assumption was met for all regression models (see Table 9 and Table 10).

Table 9

Collinearity Statistics: Individual Temperament Characteristics and Outcomes Regression Models

Independent Variables	Outcomes	Tolerance	VIF
Approach		.97	1.03
Adaptability	Total Behavior	.96	1.04
Intensity	Disruptive/Antisocial	.90	1.11
Mood	Anxiety	.94	1.06

### Table 10

Collinearity Statistics: Combination of Temperament Characteristics and Outcomes Regression Models

Independent Variables	Outcomes	Tolerance	VIF
Approach		.84	1.20
Adaptability	Total Behavior	.67	1.49
Intensity	Disruptive/Antisocial	.83	1.20
Mood	Anxiety	.65	1.54

Analysis. After the assumptions were tested and met, block one of the

hierarchical regressions comprised factors that were controlled for: cognitive ability

(estimated IQ), gender, and maternal education. Notably, given that maternal education is categorical with more than two categories, categorical control variables were created ("dummy variables") to use within the model. The second block of the regression models included the individual temperament factors of either adaptability, intensity, mood or approach, respectively, and to test the final hypothesis, all four temperament characteristics. The outcome variables examined were overall maladaptive behavior (Total Behavior Problem), disruptive/antisocial behavior, and anxiety (Tabachnick & Fidell, 2013). Tables 11 and 12 (below) summarize the results of the hierarchical regressions. Additionally, Figures 2 and 3 (below) summarize the total variance explained by the control and temperament variables. Full information for each regression is provided in Appendix A.

*Hypothesis 1.* In order to test the first hypothesis regarding whether the temperament characteristic of adaptability significantly predicting overall, internalizing, and externalizing maladaptive behavior in children with ASD later in childhood, three hierarchical regressions were conducted. First, a hierarchical linear regression was calculated to assess the significance of the temperament characteristic of adaptability in predicting overall maladaptive behavior. Neither block 1 nor block 2 incremental R<sup>2</sup> was statistically significant. The full regression model was also not statistically significant (F(5, 65) = .93, p = .47,  $R^2 = .07$ ). The beta coefficient for adaptability was also not statistically significant (p = .36). Additionally, the predictive relationship between adaptability and disruptive/antisocial behavior was statistically nonsignificant (F(5, 65) = .73, p = .60,  $R^2 = .05$ ). Neither block 1 nor block 2 incremental  $R^2$  was statistically

significant. The beta coefficient for adaptability was not statistically significant (p=.18). Finally, a third regression to examine the predictive relationship of adaptability and anxiety yielded results that were not statistically significant ( $F(5, 65) = .94, p = .46, R^2 = .07$ ). Neither block 1 nor block 2 incremental R<sup>2</sup> was statistically significant. The beta coefficient was also not statistically significant (p = .94). Further, the control variables of maternal education, gender, and cognitive ability were not significant predictors of overall maladaptive behavior or disruptive/antisocial behavior. However, for the outcome of anxiety, cognitive ability (estimated IQ) was a statistically significant predictor (p= .04).

*Hypothesis* 2. In order to test the hypothesis that overall maladaptive behavior, disruptive/antisocial behavior, and anxiety are significantly predicted by the temperament characteristic of intensity, three hierarchical regressions were conducted. The regression of overall maladaptive behavior on intensity was not statistically significant (F(5, 65) = 1.52, p = .20,  $R^2 = .10$ ) and neither block 1 nor block 2 incremental  $R^2$  was statistically significant. The beta coefficient for intensity was also not significant (p=.06) Second, the overall model representing the predictive relationship between intensity and disruptive/antisocial behavior was also not statistically significant (F(5, 65) = 2.14, p=.07,  $R^2 = .14$ ). The block 1 incremental  $R^2$  was not statistically significant, however, the block 2 incremental  $R^2$  was statistically significant (p < .001). The beta coefficient for intensity was not statistically significant (r(5, 65) = 2.14, p=.07,  $R^2 = .14$ ). The block 1 incremental  $R^2$  was not statistically significant, however, the block 2 incremental  $R^2$  was statistically significant (p < .001). The beta coefficient for intensity and anxiety was not statistically significant (F(5, 65) = 1.18, p = .33,  $R^2 = .08$ ), and neither the block 1 nor block 2 incremental  $R^2$  were statistically significant.

The beta coefficient for intensity was also not statistically significant (p=.29). However, the beta coefficient for cognitive functioning (estimated IQ) was significant with both block 1 and block 2 (p=.04 for both blocks).

Hypothesis 3. In order to test the third hypothesis that internalizing, externalizing, maladaptive behavior in later childhood in children with ASD is significantly predicted by the temperament characteristic of mood, three hierarchical regressions were again conducted. First, the predictive relationship between the temperament characteristic of mood and overall maladaptive behavior was found to be statistically significant (F(5, 65)) = 2.96, p = .02,  $R^2 = .19$ ). The incremental  $R^2$  for block 1 was not statistically significant but block 2 was statistically significant (p < .001). The beta coefficient for mood was also significant (p < .001). Additionally, the beta coefficient for cognitive functioning (estimated IQ) was significant within block 2 (p=.03). Further, the relationship between the temperament construct of mood and disruptive/antisocial behavior was found to be not statistically significant within the overall model ( $F(5, 65) = 2.22, p = .06, R^2 = .15$ ). The block 1 incremental  $R^2$  is not statistically significant, however block 2 was statistically significant (p < .001). The beta coefficient for mood was also statistically significant (p < .001). Finally, the regression model of anxiety on mood was not statistically significant (F(5, 65) = 1.84, p = .12,  $R^2 = .12$ ). The block 1 incremental  $R^2$  is not statistically significant, however block 2 was statistically significant (p = .05). The beta coefficient of mood was also statistically significant (p = .05).

*Hypothesis 4*. In order to test the fourth hypothesis that overall, internalizing, and externalizing maladaptive behavior in later childhood in children with ASD is statistically

significantly predicted by the temperament characteristic of approach, three hierarchical regressions were conducted. First, the regression of maladaptive behavior on approach yielded nonsignificant results ( $F(5, 65) = .77, p = .58, R^2 = .06$ ) and neither block 1 nor block 2 incremental  $R^2$  was statistically significant. The beta coefficient for approach was not statistically significant. Secondly, the relationship between approach and disruptive/antisocial behavior was not statistically significant ( $F(5, 65) = .36, p = .88, R^2 = .03$ ) and neither block 1 nor block 2 incremental  $R^2$  was statistically significant. The beta coefficient for approach and disruptive/antisocial behavior was not statistically significant ( $F(5, 65) = .36, p = .88, R^2 = .03$ ) and neither block 1 nor block 2 incremental  $R^2$  was statistically significant. The beta coefficient for approach was also not statistically significant (p = .79). Finally, the regression model representing the predictive relationship between the temperament characteristic of approach and anxiety was also not statistically significant ( $F(5, 65) = 1.98, p = .09, R^2 = .13$ ). The block 1 incremental  $R^2$  was not statistically significant, however block 2 was statistically significant (p = .03). The beta coefficients for approach and cognitive ability (estimated IQ) were also significant (p = .03 and p = .04 respectively).

*Hypothesis* 5. Finally, the fifth hypothesis of a combination of the temperament characteristics of approach, adaptability, intensity, and mood significantly predicting overall, internalizing, and externalizing maladaptive behavior in children with ASD later in childhood, three hierarchical regressions were calculated. Specifically, the predictive relationship between a combination of approach, adaptability, intensity, and mood and overall maladaptive behavior yielded a statistically significant regression model (*F* (8, ) =2.15, p = .04,  $R^2$  = .22) with a statistically significant incremental  $R^2$  for block 2 (p = .02). The incremental  $R^2$  for block 1 was not significant. The slopes for mood and

cognitive ability were statistically significant (p = .01 and p = .04 respectively). Further, the combination of temperament characteristics also significantly predicted the outcome of disruptive/antisocial behavior (F(8, 62) = 2.30, p = .03,  $R^2 = .23$ ) with a statistically significant incremental  $R^2$  for block 2 (p = .01). The incremental  $R^2$  for block 1 was not significant. There were statistically significant slopes for intensity and mood (p = .02 and p = .03 respectively). Finally, the predictive relationship between the combination of temperament characteristics and anxiety yielded a statistically significant model as well (F(8, 62) = 2.21, p = .04,  $R^2 = .22$ ) with a significant incremental  $R^2$  for block 2 (p = .02). The incremental  $R^2$  for block 1 was not significant. Further, the beta coefficients for approach and mood were statistically significant (p = .02 and p = .05 respectively).

Table 11 Regression Model Summaries: Individual Temperament Characteristics and Behavior Outcomes (n=71)

		Total Problem						Disruptive/Antisocial				Anxiety						
			В	enav	10r													
		<u>R<sup>2</sup></u>	$\Delta R^2$	F	<u>p</u> <u>value</u>	<u>p</u> <u>value</u> ΔF	<u>R<sup>2</sup></u>	$\Delta R^2$	<u>F</u>	<u>P</u> <u>value</u>	<u>μ</u> <u>value</u> <u>ΔF</u>	<u>R<sup>2</sup></u>	$\Delta R^2$	<u>F</u>	<u>p</u> <u>value</u>	<u>p</u> <u>value</u> ΔF		
Control Variables	Block 1	.05	.05	.95	.44	.44	.03	.03	.44	.78	.78	.07	.07	1.19	.32	.32		
Adapt.		.07	.01	.93	.47	.36	.05	.03	.73	.60	.18	.07	.00	.94	.46	.94		
Intensity	ζ2	.10	.05	1.52	.20	.06	.14	.12	2.14	.07	.004**	.08	.02	1.18	.33	.29		
Mood	Block	.19	.13	2.96	.02*	.002 **	.15	.12	2.22	.06	.004**	.12	.06	1.84	.12	.05*		
Approach		.06	.001	.77	.58	.77	.03	.001	.36	.88	.79	.13	.07	1.98	.09	.03*		

Notes:  $p \text{ value } \Delta F$ = incremental R<sup>2</sup>; \*p < 0.05 (2-tailed) ; \*\*p < 0.01 (2-tailed)

## Table 12.

Regression Model Summa	aries: Combinatio	n of Temperament	Characteristics and	d Behavior
<i>Outcomes</i> $(n=71)$				

		Total Problem Behavior				D	isrup	tive/A	ntiso	cial			Anxie	ety		
		<u>R2</u>	$\Delta R^2$	F	p <u>valu</u> <u>e</u>	<u>p</u> <u>valu</u> <u>e ΔF</u>	<u>R2</u>	$\Delta R^2$	<u>F</u>	p <u>valu</u> <u>e</u>	<u>p</u> <u>valu</u> <u>e ΔF</u>	<u>R2</u>	$\Delta R^2$	<u>F</u>	<u>p</u> <u>valu</u> <u>e</u>	<u>p</u> <u>value</u> ΔF
<b>Control Variables</b>	Block 1	.05	.05	.95	.44	.44	.0 3	.03	.44	.78	.78	.07	.07	1.19	.32	.32
Control Variables +Approach+ Adapt.+ Intensity+ Mood	Block 2	.22	.16	2.15	.04*	.02*	.2 3	.20	2.30	.03*	.01* *	.22	.15	2.21	.04*	.02*

Notes: *p* value  $\Delta F$ = incremental R<sup>2</sup>; \**p* < 0.05 (2-tailed) ; \*\**p* < 0.01 (2-tailed)

# Summary

In summary, Pearson correlations showed statistically significant correlations between the independent variables of approach and anxiety, intensity and disruptive/antisocial behavior, mood and overall maladaptive behavior, mood and disruptive/antisocial behavior, as well as mood and anxiety. Additionally, there were statistically significant correlations found between the control variable of cognitive ability (estimated IQ) and anxiety. Hierarchical multiple regressions were used to explore the predictive relationships between the individual temperament characteristics first and then the characteristics in combination. As a result, there were significant predictive relationships found between the temperament characteristic of mood and overall maladaptive behavior, disruptive/antisocial behavior, and anxiety respectively. The temperament construct of intensity was a statistically significant predictor of disruptive/antisocial behavior. Additionally, the temperament characteristics in combination yielded significant results for mood and overall maladaptive behavior. The control variable of cognitive ability (estimated IQ) was also found to be a statistically significant predictor of overall maladaptive behavior. Further, the temperament characteristics of intensity and mood were found to be statistically significant predictors of disruptive/antisocial behavior. Finally, the temperament characteristics of approach and mood were significant predictors of anxiety when the combination of temperament characteristics were examined.



Figure 2. Incremental R<sup>2</sup> of Control and Temperament Variables by Behavior Outcomes



*Figure 3.* Incremental  $R^2$  of Control and the Combination of Temperament variables by Behavior Outcomes

#### **Chapter 5: Discussion**

# Introduction

One of the most hopeful components of the differential susceptibility theory is that individuals who are highly sensitive can be particularly vulnerable, but also particularly susceptible to the positive effects of intervention (Belsky & Pluess, 2009; Lionetti et al., 2018). Similarly, research has indicated that if it is established that children have a particularly sensitive temperament or a "biologically reactive phenotype," they benefit more completely from support within their environments (Ellis, Essex, & Boyce, 2005. p. 305). Therefore, if characteristics of what constitutes a highly sensitive or reactive individual are explored more thoroughly, particularly in under-researched populations such as children with ASD, then intervention teams may be able to optimize the supports within the individual's environments and program more holistically for the child.

Overall, the results of the current study are consistent with existing research and suggest that the temperament characteristics of mood, intensity, and approach, which are often traits that are explored when looking at high sensitivity or reactivity in children, are correlated and predictive of maladaptive behavior later in life within a sample of children with ASD. Maladaptive behavior is a particularly salient negative outcome to understand for children with ASD as it has an impact on children and families' quality of life above

and beyond the skill deficits related to ASD symptomology alone (Hartley, Sikora, and McCoy, 2008; Hastings et al., 2005; Lecavalier, Leone, & Wiltz 2006).

The current research was a valuable opportunity to access rich, archival data from a longitudinal study that was conducted with children with ASD, to explore the relationships between specific temperament characteristics and later behavioral outcomes. While there were limitations to the current study, there were important implications for the field and future research that are explored below.

#### **Summary of Results**

In summary, in partial support for research question 1, hypothesis 1, showed statistically significant correlations were found between the temperament characteristics of approach and anxiety, intensity and disruptive/antisocial behavior, mood and overall maladaptive behavior, mood and disruptive/antisocial behavior, and mood and anxiety.

Hierarchical linear regressions were used to explore the predictive relationships between the individual temperament characteristics and the characteristics in combination. Consistent with research question 1, hypothesis 3, there were significant predictive relationships found between the temperament characteristic of mood and overall maladaptive behavior, disruptive/antisocial behavior, and anxiety respectively. In partial support for hypothesis 2, the temperament construct of intensity was a statistically significant predictor of disruptive/antisocial behavior. Additionally, consistent with hypothesis 5, the temperament characteristics in combination yielded significant results for mood, intensity, approach, and adaptability.

#### **Limitations of the Study**

**Diagnostic Criteria used within the Study.** The current definition of Autism Spectrum Disorder (ASD) is found within the Diagnostic Statistical Manual, Fifth Edition (DSM-5), which was released in May of 2013 (Kim et al., 2014). However due to the timeframe of the longitudinal study, the current study used the DSM-IV-TR (American Psychiatric Association, 1994) which differentiates 'Autistic Disorder,' Asperger Disorder,' ' pervasive developmental disorder not otherwise specified,' (PDD-NOS) under the broader category of Pervasive Developmental Disorders (Kita & Hosokawa, 2011).

There are studies that found that the majority of children diagnosed with diagnosed with autism, Asperger disorder, or PDD-NOS under the DSM-IV criteria meet the DSM-V criteria for Autism Spectrum Disorder (ASD) (Kim et al, 2014). Individuals with a previous diagnosis of PDD-NOS are the most likely to receive a diagnosis of Social Communication Disorder (SCD) (Kim et al., 2014). Notably, consistent with existing literature, the researcher also confirmed with the principal investigator of the longitudinal study Dr. Susan Hepburn that each individual participant within the sample met the DSM-V criteria for ASD. Due to the funding of the project, researchers were required to confirm and update the diagnosis of each participant when the DSM-V criteria were released (Dr. S. Hepburn, personal communication, 3/29/2019).

**Sample Size.** Longitudinal studies have the conceptual benefit of looking at developmental relationships over time. There are some researchers who suggest that

longitudinal studies are the best way to study populations of children with neurodevelopmental disabilities such as ASD. However, despite the benefits of longitudinal study design, the resources needed to conduct a long-term study can be difficult to maintain. Further, expecting families who have a least one child with special needs to participate in research for multiples years is often unrealistic. Thus, attrition is often high and maintaining a large sample size is a common problem, particularly for populations of children with neurodevelopmental disabilities. Consistent with other longitudinal studies, the current study was underpowered, and thus, the analyses were not as robust as they would be if there were more participants (Szatmari et al., 2017). A small sample size is often a "fatal flaw" in longitudinal design and can limit the generalizability of the research results (Szatmari et al., 2017, p.12). In order to address the limited sample size, careful consideration was taken regarding the inclusion criteria and power analyses. For example, when an outlier was identified, the pros and cons of removing one participant were weighed and the decisions on whether to remove an outlier were deliberate. Analyses were also run with and without any significant outliers identified in order understand how the outlier impacted the results. If there was significant impact, the outlier was removed (e.g. if a relationship was significant with the outlier included versus no include, the outlier was often removed).

Another secondary limitation that likely resulted from a limited sample size was that the expected relationships between the control variables of maternal education, estimated cognitive ability, and gender and later maladaptive behavior were not established. Within the existing literature, there are recognized relationships between cognitive ability, maternal education, and gender that were not found within the current study. Interesting, there were significant relationships found between estimated cognitive ability and the outcomes of overall maladaptive behavior as well as anxiety, but only when it was combined with the temperament characteristics of mood and intensity. Recommended next steps would be to explore this predictive relationship within a larger sample and see if the relationships were similar. It would also be important to see if there was any moderation or mediation. Further, critical next steps in the research would also be to understand if the small sample size was the only reason for not finding significant relationships between the control variables and the outcomes.

Lack of Environmental Variables. Both the differential susceptibility models that form the philosophical foundation for the study include an element of environmental interaction that would need to be explored in next steps within the research to establish whether the relationships identified within the study have any moderating or mediating variables. Notably, within the archival data set there were not consistent environmental variables to explore. Since the longitudinal study began, outcome research has evolved and there is a deeper understanding of the complexities of theories like the differential susceptibility model. An extensive amount of research has been conducted on the importance of factors like interventions received, parental stress, and quality of life in addition to individual child characteristics. Further, best practice within temperament research is to include environmental variables to examine how they interact with the biological aspects of temperament. Temperament researchers have evolved from looking at linear relationships to focusing on the interaction of the complicated set of variables that often influence outcomes (Belsky, Hsieh, & Crnic, 1998; Calkins, 2012).

Individual variables influencing Outcomes. Factors such as autism severity, adaptive skills, more comprehensive measures of cognitive ability, and social skills have all been found to influence outcomes for individuals with ASD (Levy & Perry, 2011). While some of this information was available within the archival data set, it was not consistently collected or entered within the master data set, and thus was not available for the current study. An important next step using the results of the current study would be to broaden the individual factors explored to see if we can understand the functioning of the current sample and if other factors influenced outcomes.

**Heterogeneity of ASD.** A known and recognized challenge when studying children with ASD is the heterogeneity in which the condition presents within each individual (Szatmari et al., 2017). Given that the nature of the diagnostic criteria is now dimensional, the presentation of what is considered "ASD" is broader than ever. Thus, consistent with this trend, the sample within the current study had a wide range of functioning that may ultimately impact how the results are interpreted.

**Homogeneity of the Sample.** An important limitation of the current study, and many studies utilizing longitudinal research, is that the study sample is not representative of the population (Szatmari et al., 2017). Specifically, the sample is mostly White and educated. This factor alone could have influenced the relationships of the control variables to the outcomes. The homogeneity of the sample also impacts who the results can generalize to as well as how we interpret the results in a culturally competent way. It is critical that future studies examine barriers to the participation within research for all families and that more diverse samples are sought to better understand these concepts and relationships comprehensively.

# Summary

In summary, the limitations within the current study include an older definition of ASD, limited sample size, lack of environmental variables, limited individual variables, the heterogeneity of how ASD presents, and the homogeneity of the sample. It should be noted that the limitations were addressed by the researcher to the best of her ability.

#### **Specific Implications**

Prevention science suggests that if risk factors are identified, predictions can be made about potential stressors and protective factors that inform intervention and treatment (Coie et al., 1993). Thus, while there were limitations to the current study that limit generalizability, there are important implications that are explored below.

**Mood and Intensity.** Within the current study, mood was found to be a significant predictor of later maladaptive behavior, disruptive/antisocial behavior, and as anxiety. Further, the results of the current study indicated a statistically significant relationship between intensity and externalizing behavior (or disruptive/antisocial). The terms of reactivity, irritability, and negative emotionality are all terms related to the concepts of intensity and mood used within the study that have been shown to have strong implications for later outcomes and quality of life (Robb, 2010; Rothbart, 2012). Intensity and mood are also part of the constellation of characteristics that comprise the concept of 'difficult' or highly sensitive temperament which has also been shown to be

related to later maladaptive behavior (Thomas & Chess, 1986). Thus, the results of the current study are consistent with existing literature regarding temperament risk factors that relate to maladaptive behavior later in life. The fact that these relationships were consistent within a sample of children with ASD is a valuable contribution to the literature. Next steps would be to conduct a similar study with a larger sample to establish if the results remain consistent within a study that has sufficient power.

Implications for intervention include early intervention that involves direct teaching of skills such as emotional regulation and self-regulation. Parents would also benefit from early coaching around effective parenting strategies for children with a 'negative' mood or who have high reactivity, particularly if they have had measures to show that they have a high level of parental stress (Sharma, Gonda, & Tarazi, 2018). Additionally, focusing on distress tolerance in multiple settings for children who demonstrate a more highly sensitive temperament may also be beneficial.

**Approach.** Also consistent with existing literature, a significant relationship was found between the temperament characteristic of approach and the development of later anxious behavior in a sample of children with ASD. Establishing that this relationship between approach and anxiety remains consistent within a sample of children with ASD is a valuable first step in understanding how characteristics associated with 'approach' such as a need for sameness, social anxiety, or difficulty with novelty can impact outcomes. Implications for intervention include teaching skills around cognitive flexibility and gradual desensitization to experiencing new activities, environments, and people could be helpful for children with ASD.

#### **Ideas for Future Research: Broader Implications**

Given the limitations of the study, the generalizability of the results were limited. As mentioned above, first steps suggested for future research and establishing the direct implications of the results of the study would be to replicate the study the analyses with a larger sample size. However, despite the limitations, the results of the current study had important implications for the field and next steps.

**Measurement of Temperament.** One of the common challenges when studying temperament is the lack of consensus around the definition to temperament. As discussed within the literature review, when studying temperament, a researcher must pick a measure that is tied to a specific theory with specific concepts and terminology around temperament characteristics. Thus, a helpful research study would be to look at the commonalities of temperament terms and concepts across measures and theories. For example, understanding how the current results related to mood and intensity interface with existing literature on related terms such as reactivity and irritability would be helpful in understanding implications of the research results.

Further, with the clarification of temperament as a construct comes the need for updated measurement. Like any construct, there needs to be measures of temperament with updated norms that are culturally relevant, reliable, and valid. There is a need for the existing measures of temperament in particular to be updated to reflect the current literature so that the most accurate information regarding the risk factor of temperament can be obtained (Frick, 2004).
**Methodology.** In order to gather richer picture of how temperament characteristics impact outcomes a mixed methods approach may be helpful. Gaining qualitative information in conjunction with quantitative information may be particularly helpful. A mixed methods approach to temperament research can also help us gain a more complete picture of existing predictive relationships and potentially establish new ones. A mixed methods approach may also be a valuable way to gain consensus regarding temperament constructs.

Interdisciplinary approach. Another important aspect that came up within the current study's literature review was the wide array of disciplines that have explored temperament, however there has been very little collaboration between disciplines on this topic. There has been a heavy medical, clinical, and genetic focus on temperament. However, school psychology is an important example of a more applied discipline that would benefit from exploring how an individual risk factor like temperament can inform school-based intervention, learning styles, school engagement, and access to learning. There has been limited research within the school psychology literature about temperament and school functioning and outcomes. Being able to provide teachers and school teams anticipatory guidance around what to expect regarding a student's behavior and functioning based off of a comprehensive assessment that includes temperament could be incredibly valuable. Often, teachers and parents refer to what they know about temperament in casual, anecdotal ways – they can make causal connections due to a child

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being "shy" or "sensitive." It would be a valuable contribution to the literature to examine more closely through research temperament characteristics and how they related to a child's educational outcomes. Additionally, examining how measuring temperament can help with interventions and programming for students with disabilities would also be helpful.

**Demonstration of Skills versus Presence of Problems.** Notably, Belsky and Pluess (2009) caution that an overrepresentation of risk factors can occur if the absence of adversity is considered as the presence of skills. Thus, an important factor to explore would be the presence of strengths and skills within populations of children with ASD. Including a broader array of individual factors, environmental variables, and a measure of skill development are all elements that should be included in future research and would contribute to a greater understanding of predictive relationships and where we can focus intervention.

**Temperament and Autism Spectrum Disorder (ASD).** While the current research supports the idea that temperament can lend valuable information regarding what individual factors might be most salient for outcomes for children with ASD, the results are preliminary. First, we must continue to clarify our understanding of the definition of temperament and refine the measurement of the construct. Then, understanding how temperament is unique from the characteristics of ASD will be critical in truly understanding the importance of temperament as a unique individual factor that influences outcomes. Further, studies that include children with ASD and other

comparison populations (e.g. typical developing and other disabilities) can be helpful in clarifying if and how the results apply uniquely to individuals with ASD.

**Resiliency.** Families with children with ASD are at a greater risk for stress and negative outcomes as families with children who are typically developing (Green, 2007; Leone, Dorstyn, & Ward, 2016; Stainton & Besser, 1998). More research is needed to explore the complex relationships between risk factor such as temperament and behavioral outcomes, particularly in special populations such as children with ASD. Next steps for research would be to explore how temperament contributes to positive outcomes such as resiliency, quality of life, and presence of skills/strengths. Additionally, exploring more the differential susceptibility theories regarding how sensitivity also relates to plasticity and receptiveness to intervention will be critical. **Conclusion** 

While many researchers have concluded that temperament is predictive of clear outcomes (Zentner & Bates, 2008), an important reminder from researcher Rothbart (2012), is "temperament is not destiny," (p.5). It is important to continue to explore early risk factors that have the potential to influence later behavioral outcomes for children with ASD so that we better understand these complex predictive relationships. With increased knowledge about the nature of what specific temperament risk factors may be the most impactful later on for individuals with ASD, there can be more effective, targeted family, child, school, and clinical interventions that can foster resiliency. Children with ASD are often considered particularly sensitive, and while these preliminary results support this notion, the current study is only a first step in establishing a broader body of literature regarding children with ASD and what promotes more positive future outcomes for the individual child and their families.

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# **Appendix A: Regression Model Coefficient Tables**

## Table 1

*Hierarchical Regression of Total Problem Behavior on Maternal Education, Gender, Cognitive Ability and Adaptability* (n=71)

В	SE B	р	Model $\Delta R^2$
		1	
			0.5
6.49	8.16	.43	
.28	6.50	.97	
2.90	7.24	.69	
18	.12	.11	
			.01
6.38	8.17	.44	
.80	6.54	.90	
2.76	7.25	.71	
20	.12	.09	
3.49	3.77	.36	
	6.49 .28 2.90 18 6.38 .80 2.76 20 3.49	B         SE B           6.49         8.16           .28         6.50           2.90         7.24          18         .12           6.38         8.17           .80         6.54           2.76         7.25          20         .12           3.49         3.77	B         SE B $p$ 6.49         8.16         .43           .28         6.50         .97           2.90         7.24         .69 $\cdot.18$ .12         .11           6.38         8.17         .44           .80         6.54         .90           2.76         7.25         .71 $20$ .12         .09           3.49         3.77         .36

	В	SE B	р	Model $\Delta R^2$
Block 1.				0.3
Maternal Education 1	.25	2.92	.93	
Maternal Education 2	-2.77	2.33	.24	
Gender	12	2.59	.97	
Cognitive Abilities	01	.04	.86	
(Estimated IQ)				
Block 2.				.03
Maternal Education 1	.19	2.90	.95	
Maternal Education 2	-2.50	2.32	.29	
Gender	19	2.57	.94	
<b>Cognitive</b> Abilities	02	.04	.67	
(Estimated IQ)				
Adaptability	1.83	1.34	.18	

*Hierarchical Regression of Disruptive Antisocial Behavior on Maternal Education, Gender, Cognitive Ability and Adaptability* (n=71)

\*p < 0.05 (2-tailed) ; \*\*p < 0.01 (2-tailed)

### Table 3

*Hierarchical Regression of Anxiety on Maternal Education, Gender, Cognitive Ability and Adaptability* (n=71)

	В	SE B	р	Model $\Delta R^2$
Block 1.				0.7
Maternal Education 1	.41	1.16	.97	
Maternal Education 2	.06	.93	.95	
Gender	13	1.03	.90	
Cognitive Abilities	.04	.02	.04*	
(Estimated IQ)				
Block 2.				.00
Maternal Education 1	.04	1.17	.97	
Maternal Education 2	.05	.94	.96	
Gender	13	1.04	.90	
Cognitive Abilities	.04	.02	.04*	
(Estimated IQ)				
Adaptability	04	.54	.94	

	В	SE B	р	Model $\Delta R^2$
Block 1.				0.5
Maternal Education 1	6.49	8.16	.43	
Maternal Education 2	.28	6.50	.97	
Gender	2.90	7.24	.69	
Cognitive Abilities	18	.12	.11	
(Estimated IQ)				
Block 2.				.05
Maternal Education 1	11.28	8.39	.18	
Maternal Education 2	1.52	6.41	.81	
Gender	3.51	7.10	.62	
Cognitive Abilities	20	.11	.08	
(Estimated IQ)				
Intensity	8.07	4.23	.06	
-				

*Hierarchical Regression of Total Problem Behavior on Maternal Education, Gender, Cognitive Ability and Intensity* (n=71)

\*p < 0.05 (2-tailed); \*\*p < 0.01 (2-tailed)

### Table 5

*Hierarchical Regression of Disruptive/Antisocial Behavior on Maternal Education, Gender, Cognitive Ability and Intensity* (n=71)

D	SE B	р	Model $\Delta R^2$
			0.3
.25	2.92	.93	
-2.77	2.33	.24	
12	2.59	.97	
-	.04	.86	
.01			
			.12
2.82	2.90	.34	
	2.21	.34	
1.11			
.21	2.45	93	
02	.04	.70	
4.32	1.46	.004**	
	.25 -2.77 12 .01 2.82 1.11 .21 02 4.32	$\begin{array}{cccccccccccccccccccccccccccccccccccc$	$\begin{array}{cccccccccccccccccccccccccccccccccccc$

	В	SE B	р	Model $\Delta R^2$
Block 1.				0.7
Maternal Education 1	.04	1.16	.97	
Maternal Education 2	.06	.93	.95	
Gender	13	1.03	.90	
Cognitive Abilities	.04	.02	.03*	
(Estimated IQ)				
Block 2.				.02
Maternal Education 1	.43	1.22	.73	
Maternal Education 2	.16	.93	.87	
Gender	08	1.03	.94	
Cognitive Abilities	.03	.02	.04*	
(Estimated IQ)				
Intensity	.65	.61	.29	

*Hierarchical Regression of Anxiety on Maternal Education, Gender, Cognitive Ability and Intensity* (n=71)

\*p < 0.05 (2-tailed) ; \*\*p < 0.01 (2-tailed)

#### Table 7

Hierarchical Regression of Total Problem Behavior on Maternal Education, Gender, Cognitive Ability and Mood (n=71)

	В	SE B	р	Model $\Delta R^2$
Block 1.				0.5
Maternal Education 1	6.49	8.16	.43	
Maternal Education 2	.28	6.50	.97	
Gender	2.90	7.24	.69	
Cognitive Abilities	18	.12	.11	
(Estimated IQ)				
Block 2.				.13
Maternal Education 1	7.70	7.64	.32	
Maternal Education 2	-2.82	6.16	.65	
Gender	-1.11	6.88	.87	
Cognitive Abilities	24	.11	.03*	
(Estimated IQ)				
Mood	10.93	3.38		
			.002**	

	В	SE B	р	Model $\Delta R^2$
Block 1.				.03
Maternal Education 1	.25	2.92	.93	
Maternal Education 2	-2.77	2.33	.24	
Gender	12	2.59	.97	
<b>Cognitive Abilities</b>	01	.04	.86	
(Estimated IQ)				
Block 2.				.12
Maternal Education 1	.66	2.76	.81	
Maternal Education 2	-3.82	2.22	.09	
Gender	-1.47	2.48	.56	
Cognitive Abilities	03	.04	.50	
(Estimated IQ)				
Mood	3.69	1.22	.004**	

*Hierarchical Regression of Disruptive/Antisocial Behavior on Maternal Education, Gender, Cognitive Ability and Mood* (n=71)

\*p < 0.05 (2-tailed) ; \*\*p < 0.01 (2-tailed)

### Table 9

*Hierarchical Regression of Anxiety on Maternal Education, Gender, Cognitive Ability* and Mood (n=71)

	В	SE B	р	Model $\Delta R^2$
Block 1.				.07
Maternal Education 1	.04	1.16	.97	
Maternal Education 2	.06	.93	.95	
Gender	13	1.03	.90	
Cognitive Abilities		.02	.04*	
(Estimated IQ)	.04			
Block 2.				.06
Maternal Education 1	.16	1.14	.89	
Maternal Education 2	24	.92	.80	
Gender	51	1.02	.62	
Cognitive Abilities	.03	.02	.07	
(Estimated IQ)				
Mood	1.03	.50	.05*	

	В	SE B	р	Model $\Delta R^2$
Block 1.				.05
Maternal Education 1	6.49	8.16	.43	
Maternal Education 2	.28	6.50	.97	
Gender	2.90	7.24	.69	
Cognitive Abilities	18	.12	.11	
(Estimated IQ)				
Block 2.				.001
Maternal Education 1	6.60	8.23	.43	
Maternal Education 2	.37	6.56	.96	
Gender	2.88	7.29	.70	
Cognitive Abilities	19	.12	.11	
(Estimated IQ)				
Approach	.82	2.78	.77	

*Hierarchical Regression of Total Problem Behavior on Maternal Education, Gender, Cognitive Ability and Approach* (n=71)

\*p < 0.05 (2-tailed) ; \*\*p < 0.01 (2-tailed)

#### Table 11

*Hierarchical Regression of Disruptive/Antisocial on Maternal Education, Gender, Cognitive Ability and Approach* (n=71)

	В	SE B	р	Model $\Delta R^2$
			^	
Block 1.				.03
Maternal Education 1	.25	2.92	.93	
Maternal Education 2	- 2.77	2.33	.24	
Gender	12	2.59	.97	
Cognitive Abilities	01	.04	.86	
(Estimated IQ)				
Block 2.				.001
Maternal Education 1	.22	2.94	.94	
Maternal Education 2	-2.80	2.35	.24	
Gender	11	2.61	.97	
Cognitive Abilities	01	.04	.90	
(Estimated IQ)				
Approach	27	.99	.79	

	В	SE B	р	Model $\Delta R^2$
Block 1.				.07
Maternal Education 1	.04	1.16	.97	
Maternal Education 2	.06	.93	.95	
Gender	13	1.03	.90	
<b>Cognitive Abilities</b>	.04	.02	.04*	
(Estimated IQ)				
Block 2.				.07
Maternal Education 1	.15	1.13	.90	
Maternal Education 2	.14	.90	.87	
Gender	16	1.00	.88	
Cognitive Abilities	.03	.02	.08	
(Estimated IQ)				
Approach	.84	.38	.03*	

*Hierarchical Regression of Anxiety on Maternal Education, Gender, Cognitive Ability and Approach* (n=71)

p < 0.05 (2-tailed); p < 0.01 (2-tailed)

#### Table 13

*Hierarchical Regression of Total Problem Behavior on Maternal Education, Gender, Cognitive Ability and All Temperament Variables (n=71)* 

	В	SE B	p	Model $\Delta R^2$
Block 1.				.05
Maternal Education 1	6.49	8.16	.43	
Maternal Education 2	.28	6.50	.97	
Gender	2.90	7.24	.69	
Cognitive Abilities	18	.12	.11	
(Estimated IQ)				
Block 2.				.16
Maternal Education 1	11.24	8.07	.17	
Maternal Education 2	-2.51	6.37	.70	
Gender	74	6.96	.92	
Cognitive Abilities	24	.11	.04*	
(Estimated IQ)				
Approach	03	2.78	.99	
Adaptability	-2.80	4.20	.51	
Intensity	5.73	4.21	.18	
Mood	11.44	4.05	.01**	

	В	SE B	р	Model $\Delta R^2$
			•	
Block 1.				.03
Maternal Education 1	.25	2.92	.93	
Maternal Education 2	-2.77	2.33	.24	
Gender	12	2.59	.97	
Cognitive Abilities	01	.04	.86	
(Estimated IQ)				
Block 2.				.20
Maternal Education 1	2.59	2.82	.36	
Maternal Education 2	-3.19	2.23	.16	
Gender	-1.03	2.43	.68	
Cognitive Abilities	03	.04	.48	
(Estimated IQ)				
Approach	54	.97	.58	
Adaptability	.20	1.47	.89	
Intensity	3.48	1.47	.02*	
Mood	3.23	1.42	.03*	

Hierarchical Regression of Disruptive/Antisocial Behavior on Maternal Education, Gender, Cognitive Ability and All Temperament Variables (n=71)
# Table 15

	В	SE B	р	Model $\Delta R^2$
			-	
Block 1.				.07
Maternal Education 1	.04	1.16	.97	
Maternal Education 2	.06	.93	.95	
Gender	13	1.03	.90	
Cognitive Abilities	.04	.02	.04*	
(Estimated IQ)				
Block 2.				.15
Maternal Education 1	.73	1.15	.53	
Maternal Education 2	23	.91	.80	
Gender	49	.99	.62	
Cognitive Abilities	.03	.02	.09	
(Estimated IQ)				
Approach	.93	.40	.02*	
Adaptability	-1.06	.60	.08	
Intensity	.68	.60	.26	
Mood	1.17	.58	.05*	

Hierarchical Regression of Anxiety on Maternal Education, Gender, Cognitive Ability and All Temperament Variables (n=71)

\*p < 0.05 (2-tailed) ; \*\*p < 0.01 (2-tailed)

## **Appendix B:** Copy of IRB Approval Letter



DATE:	February 14, 2019		
TO:	Caren Rhodes, Ed.S.		
FROM:	University of Denver (DU) IRB		
PROJECT TITLE:	[731918-1] Exploring early temperament predictors of maladaptive behavior in middle childhood in a group of children with Autism Spectrum Disorder		
SUBMISSION TYPE:	New Project		
ACTION:	EXEMPTION GRANTED		
DECISION DATE: EXEMPTION VALID THR February 13, 2021	February 14, 2019 OUGH:		
NEXT REPORT DUE: RISK LEVEL:	February 13, 2021 Minimal Risk		
<b>REVIEW CATEGORY:</b>	Exemption category # 4		

Exemption 4: Secondary Research for Which consent is Not Required

Research in this category covers secondary research uses of identifiable private information or identifiable biospecimens if at least one of the following criteria is met:

- 1. Information or biospecimens are publicly available
- The recorded information cannot readily be identified (directly or indirectly/linked); investigator does not contact subjects and will not re- identify the subjects
- Information collection and analysis involving identifiable health information when use is regulated by HIPAA "health care operations" or "research" or "public health activities and purposes".

1. Research by or on behalf of Federal department/agency using government –generated or collected information compliant with relevant privacy protections.

**Identifiable private information** is private information for which the identity of the subject is or may readily be ascertained by the investigator or associated with the information.

An **identifiable biospecimen** is a biospecimen for which the identity of the subject is or may readily be ascertained by the investigator or assisted with the biospecimen

Thank you for your submission of Exemption materials for this project. The University of Denver IRB has determined this project is **EXEMPT FROM IRB REVIEW** according to federal regulations. This exemption was granted based on appropriate criteria for granting an exemption and a study design wherein the risks have been minimized.

Exempt status means that the study does not vary significantly from the description that has been provided and further review in the form of filing an annual Continuing Review/Progress Report is not required.

Research Classified as Minimal Risk

Please note that maintaining exempt status requires that (a) risks of the study remain minimal; (b) that anonymity or confidentiality of participants, or protection of participants against any increased risk due to the internal knowledge or disclosure of identity by the researcher, is maintained as described in the application; (c) that no deception is introduced, such as reducing the accuracy or specificity of information about the research protocol that is given to prospective participants; (d) the research purpose, sponsor, and recruited study population remain as described; and (e) the principal investigator (PI) continues and is not replaced.

Implementation of Changes to Protocol or Personnel

If changes occur in any of the features of the study as described above, this may affect one or more of the conditions of exemption and may warrant a reclassification of the research protocol from exempt and require additional IRB review. For the duration of your research study, any changes, including the addition of new personnel in the proposed study, must be reviewed by the University of Denver IRB before implementing those changes.

Unanticipated Problems Involving Risks to Subjects or Others (UPIRTSOs)

Any incident, experience or outcome which has been associated with an unexpected event(s), related or possibly related to participation in this research, and suggests that the research places the subjects or others at a greater risk of harm than was previously known or suspected must be reported to the IRB.

#### **Review Period**

This exemption has been granted for a two-year time period. The DU Human Research Protection Program (HRPP)/Institutional Review Board (IRB) will retain a copy of this correspondence

within our records and will administratively close this project at the end of the two-year period unless otherwise instructed via correspondence from the Principal Investigator. Please contact the DU HRPP/IRB if the study is completed before the two-year time period or if you are no longer affiliated with the University of Denver.

Study Completion and Final Report

A Final Report is requested, via the IRBNet system, when this study has been completed. All records associated with this study must be retained in a secure location for a minimum of the three years after the completion of the project.

If you have any questions, please contact the University of Denver Human Research Protection Program/Institutional Review Board at (303) 871-2121 or at <u>IRBAdmin@du.edu</u>. Please include your project title and IRBNet number in all correspondence with the IRB; This letter has been electronically signed in accordance with all applicable regulations, and a copy is retained within the University of Denver (DU) IRB records.

### Appendix C: Secondary Data Use From: IRB Application

#### SECONDARY DATA USE FORM

Complete this form if research involves use of existing data or data being collected for non-research purposes.

Use this form, instead of the Research Narrative or Exempt Application From, if your study is limited to analysis of existing data, documents, records, or specimens.

#### NOTE:

- If your study is limited to analysis of de-identified existing data, documents, records, or specimens, it may not be necessary to complete and submit a full IRB application.
- If you have questions about whether or not your research requires review, contact <u>IRBAdmin@du.edu</u>
- Do NOT complete or submit the Research Narrative or Exempt Application Form with this form.
  If these are needed, you will be notified if a complete IRB application will be required via IRBNet.

If you have questions about filling out this form, please email <u>IRBAdmin@du.edu</u> or call (303) 871-2121.

#### 1. RESEARCH PURPOSE

1.1. Briefly explain the purpose of research, the research questions, and the potential value.

*Purpose of Research:* Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder that impacts individuals' fundamental abilities to communicate, relate, and interact socially across each stage of development. Additionally, individuals with ASD have a broad category of characteristics referred to as rigid and repetitive behaviors (American Psychiatric Association, 2013; Kim et al., 2014). Per a 2018 Center of Disease Control (CDC) report, within the United States the prevalence of ASD is 1 in 59, with males being four times more likely to be identified with ASD than females (Baio et.al, 2018). The overall prevalence of ASD is steadily increasing, with no clear reason for why.

Temperament is the biologically based behavioral style of an individual that is consistent over time (Krieger & Stringaris, 2015). Temperament influences how a person reacts and responds to novelty and adversity and ultimately influences individuals' outcomes (Carey, 1998; Zentner & Bates, 2008). The purpose of the proposed research is to explore the individual characteristic of temperament measured in early childhood and how it relates to maladaptive behavior in middle childhood in a group of children with ASD within an existing data set. The specific temperament characteristics of interest are typically associated with what the literature refers to as 'difficult' or highly sensitive temperament. There is a broad body of literature that suggests that 'difficult temperament' is defined by individuals who have the temperament characteristics of: low adaptability, negative mood, low approach, and high intensity. The existing literature suggests that individuals with highly sensitive temperament are more at risk for a broad

range of negative outcomes over time (Bates, Maslin, Frankel, 1985; Chess, 1990; Lerner & Vicary, 1984; McDevitt & Carey, 1977; Nelson, Martin, Hodge, Havill, & Kamphaus, 1999; Zentner & Bates, 2008). However, the majority of the existing temperament literature is within the typically developing population and there is not enough research to indicate whether the relationships and outcomes that are established within individuals who are typically developing are the same or differ for individuals with ASD. The consensus within the existing literature, is that children with ASD have a distinct temperament when compared to typically developing individuals as well as individuals with other neurodevelopmental disabilities. For example, by the age of one year, children with ASD can have difficulty with selfregulation (which can cause intense and more frequent distress reactions), are more irritable, have increased negative affect, they have difficulty with adapting to novel situations, and their behavioral inhibition can either be low or high as compared with children who are typically developing (Schwartz et al., 2009). Further, the existing literature on individuals with ASD and temperament suggests that temperament yields important information regarding outcomes for individuals with ASD. For example, 'difficult' or highly sensitive temperament reported within children of ASD has been shown to exacerbate individuals' social difficulties and increase the incidence of maladaptive behavior and other negative outcomes for children with ASD (Schwartz et al., 2009). The purpose of the proposed study would be to explore the prognostic value of the temperament characteristics typically associated with highly sensitive temperament and if they significantly predict maladaptive behavior within a sample of children with ASD.

**Research Questions:** There are two main research questions within the proposed study. Using existing data, the first question is broadly exploring the relationships between temperament characteristics measured within early childhood and how they are related to overall maladaptive behavior, as well as externalizing and internalizing maladaptive behavior, in middle childhood within a group of children with ASD. The second research question investigates if the specific temperament characteristics associated with highly sensitive temperament (adaptability, intensity, approach, and mood) predict maladaptive behavior such as gender and maternal education within a group of children diagnosed with ASD.

*Potential value*: Overall, the proposed study would make a novel contribution to the existing body of literature on temperament and ASD. Specifically, according to Hartley, Sikora, and McCoy (2008), clinically significant maladaptive internalizing and externalizing behavior causes greater distress to the individual and family than the primary symptoms of ASD. Thus, gathering more information about a potential individual risk factor like temperament that can lead to maladaptive behavior and other negative outcomes over time can contribute information to the field that allows professionals to give families anticipatory guidance and support after their child has received a diagnosis of ASD. Additional research regarding risk factors can also give professionals and intervention teams more information regarding how to prioritize interventions and support families. Finally, understanding the nuances of different temperament characteristics for children with ASD may help to clarify the different manifestations of the behavioral phenotype of ASD and ultimately the wide variety of outcomes for children of ASD (Schwartz et al., 2009; Hepburn & Stone, 2008).

1.2. What are the study aim(s) question(s) or hypotheses this activity is designed to answer.

**Research Questions:** There are two main research questions within the proposed study. Using existing data, the first question is broadly exploring the relationships between temperament characteristics measured within early childhood and how they are related to overall maladaptive behavior, as well as externalizing and internalizing maladaptive behavior, in middle childhood within a group of children with ASD. The second research question investigates if the specific temperament characteristics associated with highly sensitive temperament (adaptability, intensity, approach, and mood) predict maladaptive behavior above and beyond other factors that are known to be significant predictors of maladaptive behavior such as gender and maternal education within a group of children diagnosed with ASD.

*Study Hypotheses*: It is hypothesized that the specific temperament characteristics of adaptability, mood, intensity, and approach will be shown to be significantly correlated to overall, internalizing, and externalizing maladaptive behavior in middle childhood. Subsequently, it is hypothesized that the specific characteristics of adaptability, mood, intensity, and approach will significantly predict maladaptive behavior individually. It is also hypothesized that when a child with ASD has multiple characteristics associated with highly sensitive temperament (e.g. low adaptability and negative mood) that this also significantly predicts maladaptive behavior within middle childhood.

2. RESEARCH PROCEDURES INVOLVED IN THE SECONDARY DATA ANALYSIS 2.1. Provide a complete description of your study design and all the study procedures that you will perform.

For example:

- How will data be obtained?
- Are the data identifiable?
- Will any member of the study team have access to the code that links identifiers to subjects?
- Did subjects provide consent for original data?
- If a HIPPA waiver is needed to access existing private data, make this clear.

**How will the data be obtained?** The proposed study is an ex post facto (after the fact), nonexperimental design that involves secondary data analyses of data collected from previous trials of a large-scale longitudinal study conducted by researchers at JFK Partners, Center of Excellence in Autism and Neurodevelopmental Disabilities in collaboration with the University of Colorado Anschutz Medical School. The original study is entitled: Longitudinal Study of the Developing Phenotype of Autism, with principal investigator Dr. Susan Hepburn. Dr. Hepburn is also consulting as an expert advisor on the current study (please see attached Letter of Approval). Previous to Dr. Hepburn, Dr. Rogers and Pennington were the principal investigators who initiated the longitudinal study in 1996 exploring the phenotype of Autism within a group of young children as part of the Collaborative Programs of Excellence in Autism Network Projects (CPEA), funded by the National Institute of Child Health and Development (NICHD). In 2001, Dr. Rogers transferred to the University of California at Davis, to the M.I.N.D. Institute and mentored Dr. Hepburn. In 2003, the research team received funding to continue and extend the study, with Dr. Hepburn as the principal investigator. She continued the study through several more trials at JFK Partners, Center for Excellence in Autism and Neurodevelopmental Disabilities.

Are the data identifiable? No direct personal identifiers will be within the data set. Indirect personal identifiers related to the research questions such as gender, age, and maternal education level will be included.

#### Will any member of the study team have access to the code that links identifiers to subjects? No.

**Did subjects provide consent for original data?** Yes. Please see the attached for a copy of the informed consent form used within the original study. An example of a full IRB protocol submission that was approved for the original study is also also that details consent procedures and methods of human protection.

#### If a HIPPA waiver is needed to access existing private data, make this clear. No.

**Study Procedures:** The hypotheses detailed above will be analyzed using multivariate statistics including Pearson correlation and hierarchical multiple regression. The first step in the analysis process, will be preparing the data for analysis. The steps to prepare for analysis will include: addressing any missing data, running descriptive statistics, addressing outliers, and to making sure the assumptions are met for each analysis process. Next, the validity and reliability of the temperament measure will be established for the sample of children with ASD within the study. In order to address research question one regarding establishing relationships between the variables, Pearson correlation will be used to determine how strong the relationship is between the independent and dependent variables (Gliner, Morgan, & Leech, 2009; Schwartz et al., 2009). Specifically, correlations will be run to explore parent report of child temperament collected in early childhood and overall, internalizing, and externalizing behavior within middle childhood. In order to answer the second research question regarding what predicts maladaptive behavior, hierarchical multiple regressions will be used to determine if the temperament characteristics of adaptability, mood, intensity, and approach predict overall maladaptive behavior, as well as externalizing, and internalizing maladaptive behaviors.

#### 3. EXISTING DATASET

3.1. Are the data existing at the current time?



Who/what is the source from which the data set will be obtained?

#### (data should not be obtained prior to IRB approval or exemption)

The de-identified, archived data will be obtained from Dr. Hepburn. Please see attached for her letter of official approval.

# 3.2. Describe the process for gaining permission to use the data set, including any requirements, agreements, or credentials necessary to access the data. Attach a letter of cooperation or agreement for the data access.

Through the researcher's Ph.D. program she had the opportunity to participate in the Leadership Education in Neurodevelopmental Disabilities (LEND) program through JFK Partners at University of Colorado Anschutz medical campus. At this time, Dr. Susan Hepburn became the researcher's clinical and research mentor. Through this professional relationship, Dr. Hepburn also became the researcher's dissertation mentor and ultimately granted her permission to use the data from her longitudinal study within her dissertation research. Dr. Hepburn has full rights, permission to use, and ownership of the data that she obtained through JFK partners as the principal investigator of the study.

#### 3.3. Does the original file contain direct or indirect personal identifiers?



**Direct personal identifiers** include information such as: name, address, telephone number, social security number, identification number, medical record number, license number, photographs, biometric information, etc.



**Indirect personal identifiers** include information such as: race, gender, age, zip code, IP address, major, etc.





3.4. If you answered 'YES' o either, please respond to the following:

- 1. Please list the personal identifiers (direct and indirect) that will be included in the data set: Gender, Child Age, Maternal Education
- 2. Will you remove the identifiers from the data set or otherwise maintain and analyze the data in such a manner that individuals cannot be identified either directly or indirectly thorugh identifiers linked to participants? (A de-identified data set refers to original data that has been stripped of all elements that might enable a reasonably informed and determined person to deduce the identity of the participant.)



#### If answered 'NO', please provide brief justification:

Please note, the specific variables related to the research questions will not be removed. The participants within the dataset are identified using randomly assigned research numbers. There will be no direct identifiers of the participants or their families included within the data set.

3.5. Please clarify the following regarding the consent process (Choose 1 below)

Consent for use of the data for this purpose was obtained at the time of the data were originally collected. Attach the original consent document.

Consent will be obtained from each participant group.

If you plan to obtain consent from the participants, you must complete and attach either the informed consent form (usng the DU IRB Template online) or the exempt information sheet (if research is exempt).

Waiver of consent is requested.

Please complete the 'Waivers of Full Partial Consent" form. Be sure to select the option for a "Waiver or Alteration of Consent" and to provide sufficient justification in each of the boxes provided.

# Please describe briefly how consent will be obtained, or if applicable, why consent will not be obtained.

Consent will not be explicitly obtained given that the proposed study is secondary data analysis and the participants gave their fully informed consent for the original study as well as use of the data from the research in subsequent studies. It should be noted that at each time point of the longitudinal study, fully informed consent was obtained and consent data was carefully tracked and logged by the research team. The original study took careful steps to ensure that all human rights, welfare, and confidentiality were and continue to be protected. Also, due to the deidentified nature of the dataset, the researcher of the proposed study would not have any way to obtain consent directly from the participants.

#### 3.6. Explain how researchers will maintain confidentiality of the data.

Describe how researchers will protect data against disclosure to the public or to other researchers or nonresearchers. Other than members of the research team, explain who will have access to the data (e.g., sponsors, advisors, government agencies), and how long identifiable data will be kept.

The researcher has updated CITI training and a high level of training in research methods and statistics in regard to ethics and integrity within human subjects research (please see CITI training certificates attached). The researcher will keep and analyze the data electronically in a secure location on a flash drive and a backup copy on a secure, password protected hard drive that only the researcher has access to. Any capability to password protect the SPSS files, the researcher will take advantage of. She will use the technology resources at the University of Denver to make sure the data is as secure as possible. All data will be accessed through secure internet connection only, never through public Wi-Fi. All direct correspondence regarding the data (e.g. to her analysis expert on her dissertation committee) and any electronic cloud storage of the results that may need to be used will be through the University of Denver's Office 365 (e.g. OneDrive and Outlook) which is secure and HIPAA compliant. No one other than her dissertation committee and Dr. Hepburn (the owner of the data) who is an expert consultant on her dissertation research will have access to the data. The research is unfunded, so no sponsors or government agencies will have access to the data. The data may be kept for up to a year after the researcher completes her graduation requirements to ensure ample time to make any edits to her dissertation publication. After that period of time, she will destroy/delete the data in a secure manner and from all storage modalities that are within her possession (e.g. flash drive, hard drive, and cloud).

#### 3.7 Do you anticipate using these data for other studies in the future?

Yes

If answered 'YES' please explain

N/A