## Developmental Contributions of Emotion Regulation and Joint Attention in the Emergence of Autism Spectrum Disorder

by

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#### ABSTRACT

Current research efforts are focused on identifying the developmental pathways leading to the emergence of autism spectrum disorder (ASD). Prospective studies of infants at an increased likelihood of ASD report difficulties in emotional regulation (ER) and coordination of social attention (i.e., joint attention) among the earliest signs of ASD, appearing by 12 months of age. How these early-developing processes interact to predict ASD emergence is poorly understood. As such, multimodal, observational studies in infant siblings of children diagnosed with ASD are needed, as ascertainment of likelihood status does not depend on clinical ascertainment, thus an earlier developmental period can be examined. This dissertation examined the developmental relations between early ER and joint attention in infants who were at an increased likelihood (IL; have an older sibling with ASD) and low likelihood (LL; no family history of ASD) of an ASD diagnosis. Three areas were investigated: (1) the interrelations between early ER and social communication in typical development; (2) the relationship between behavioral affect and physiological arousal as measures of ER in IL and LL infants; and (3) how behavioral affect and physiological indices of ER can predict joint attention and ASD symptoms in IL and LL infants. IL and LL infants were assessed and compared at multiple time points during the first two years of life to track the development of ER and joint attention. Behavioral and physiological indices of ER were measured at 6, 12, and 18 months during an emotion-eliciting task called the Laboratory Temperament Assessment Battery. Joint attention was measured at 18 months using a modified version of the Early Social Communication Scales, and ASD symptoms were measured at 18 and 24 months. The results indicated that there was a relationship between ER and joint attention, as well as ER and ASD symptoms, among IL infants who were later classified with ASD. Moreover, joint attention was also related to ASD symptoms in both IL and

LL infants. Taken together, the findings indicated that there is a developmental relationship between ER and joint attention during infancy, which in turn influences the emergence of ASD. That is, a complex interplay between emotion dysregulation and early deficits in joint attention may influence the emergence of ASD in IL infants. Results are discussed in relation to theoretical models on the developmental link between ER and social communication skills.

#### PREFACE

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Building upon previous research and pilot data conducted, the ISS research group assisted in the concept formation of the studies conducted in Chapter 3 and Chapter 4. However, I was responsible for data collection, analysis, and manuscript composition. Chapter 1 (introduction), Chapter 2 (literature review), and Chapter 5 (concluding discussion) are my original work.

## **DEDICATION**

For my parents.

Thank you for inspiring me to pursue my dreams and giving me the strength to achieve them. I would have never been able to come this far without you.

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## **GLOSSARY OF TERMS**

ASD	Autism spectrum disorder
ER	Emotion regulation
IL	Infants at increased likelihood for ASD
LL	Infants at low likelihood for ASD
Lab-TAB	Laboratory Temperament Assessment Battery
ADOS-2-T	Autism Diagnostic Observation Schedule $-2^{nd}$ Edition $-$ Toddler Module
IJA	Initiating joint attention
RJA	Responding joint attention

#### **CHAPTER 1: Introduction**

#### 1.1. Autism Spectrum Disorder

Autism spectrum disorder (ASD) is a neurodevelopmental condition characterized by deficits in social communication, and the presence of restricted and repetitive behavior and/or interests (American Psychiatric Association, 2013). Although present from birth (Kanner, 1943; Nicolson & Szatmari, 2003), behavioral features of ASD have not been reliably detected until twelve months of age (e.g., Luyster et al., 2009; Shen & Piven, 2017). As such, identifying early risk markers of ASD that emerge during the first year of life is of critical importance. To examine the emergence of ASD during the first few years of life, researchers have utilized the prospective, infant sibling study design. Sibling recurrence risk of ASD from studies using this design is estimated at 18.7 percent (Ozonoff et al., 2011); thus, researchers can longitudinally assess the early development of infants with (i.e., at increased likelihood of ASD) and without (i.e, at low likelihood of ASD) an older sibling with ASD to capture the early emergence of ASD. These studies uncover characteristics that emerge during the first years of life to describe those infant siblings who are more likely to be diagnosed with ASD (Ozonoff, 2009; Szatmari et al., 2016). As such, this methodology is ideal for elucidating early risk markers, which may lead to improvements in early detection, diagnosis, and intervention for ASD. Furthermore, such findings may inform studies of the underlying mechanisms and developmental pathways leading to the emergence of ASD.

Previous prospective longitudinal baby sibling research has uncovered the roles of a range of behavioral and neurocognitive markers (e.g., social interaction, language, restrictive repetitive behaviors, executive functioning, and motor development (Jones et al., 2014). Very little research has explored the relationship between emotion regulation, as measured by behavioral and physiological indexes, and social communication development. As such, the present dissertation focuses on understanding the role of each and how they are related to the emergence of ASD in a cohort of infants who are at increased likelihood of an ASD diagnosis and their low likelihood peers.

To provide further rationale for the dissertation aims and predictions, the introduction will first examine the constructs of emotion and emotional regulation, social communication, and the association between emotion regulation and social communication. Next, theoretical frameworks underlying these relationships will be explored. Finally, the research questions and hypotheses guiding the dissertation will be presented.

#### 1.2. Emotion

Emotions play a critical role in guiding thoughts and actions, appraisal of experiences, and regulating behavior (Cole et al., 2004; Lehtonen et al., 2012). Despite considerable debate among theorists, there is no consensus regarding the precise definition of emotion. The present thesis work will be guided by a contemporary perspective of emotion. Among contemporary emotion theorists, it is widely agreed that emotions are a collection of psychological states that are comprised of three response components: (1) subjective experience, (2) behavioral responses (i.e., facial affect, verbal response, etc.), and (3) physiological responses (i.e., changes in heart rate, respiration, etc.). That is, as emotions arise in response to environmental stimuli, the loose coupling of experiential, behavioral, and physiological responses occur. Consequently, this enables an individual to feel, behave, and organize whole-body responses (Mauss et al., 2005; Gross & Barrett, 2011; Gross & Jazaieri, 2014). But what are the core features or conditions required for an emotion to occur? First, as outlined by Gross & Thompson (2007), emotions arise when an individual attends to a situation, appraises it, and judges it with respect to his/her goals. These goals may be transient or enduring, conscious or unconscious, peripheral to oneself or hold meaning to one's sense of self. Irrespective of the type, each goal retains significance. In fact, this significance gives rise to and determines the quality of the emotion (Campos et al., 2004). Second, as previously described, emotions involve loose coupling of subjective experience, expressive behavior, and physiological responses. The subjective aspect of emotion allows an individual to 'feel' and subsequently produces neurological impulses, resulting in the individual acting upon the emotion. Impulses are associated with central and peripheral physiological changes that both anticipate and follow the behavioral response. Third, emotions are malleable in nature and may be modulated in a number of ways. That is, emotions may interrupt one's activities and force themselves on his/her awareness (i.e., are dominant), or they may compete with other responses that are also triggered by the situation at hand. It is this third aspect of emotion that makes emotional regulation possible (Gross & Thompson, 2007).

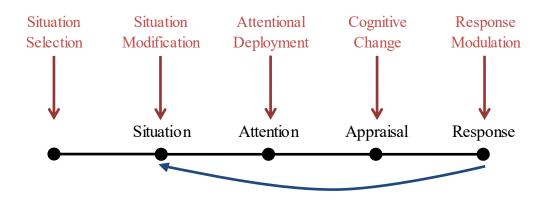
#### **1.3. Emotion Regulation**

Emotion regulation (ER) refers to "extrinsic and intrinsic processes responsible for monitoring, evaluating, and modifying emotional reactions, especially their intensive and temporal features, to accomplish one's goal" (Thompson, 1994). Emotion regulatory processes may take several forms depending on context (Gross & Jazaieri, 2014). For example, ER may be conscious or unconscious, automatic or controlled, intrinsic (i.e., regulating one's own emotions) or extrinsic (i.e., regulating another's emotions), and may have effects on one or multiple components in the emotion-generative process (Gross & Thompson, 2007). Moreover, given that emotions are comprised of multiple response components (i.e., experiential, behavioral, and physiological responses) that unfold over time, ER involves changes in the rise time, magnitude, latency, and duration of these responses (Gross & Thompson, 2007).

Despite the various forms of ER, Gross and Jazaieri (2014) outlined three core features and necessary conditions for the adaptive regulation of emotion. First, ER involves the awareness of emotions, as well as the context in which they're being expressed. That is, emotional awareness enhances not only the range of available regulatory strategies, but also the flexibility in how these strategies are utilized. Second, ER may intensify, dampen, or maintain the magnitude and/or intensity of an emotional experience/behavior/physiology depending on an individual's goals. Third, for effective and adaptive ER to occur, it is imperative that specific strategies are employed to achieve the ER goal (Gross & Jazaieri, 2014).

#### 1.3.1. The Process Model of Emotional Regulation

The Process Model of ER is a widely accepted framework for organizing emotion regulatory processes (Gross, 1998; Gross & Thompson, 2007; Gross & Jazaieri, 2014). As illustrated in Figure 1.1, the Process Model is comprised of five families of ER processes: situation selection, situation modification, attentional deployment, cognitive change, and response modulation. Situation selection involves actions that make it likely that an individual will be in a situation that he/she expects to give rise to desirable (or undesirable) emotions. Situation modification occurs when an effort is made to alter or change the situation in order to modify the emotional impact. Attentional deployment occurs when an individual attempts to direct (or re-direct) attention to alter his/her emotional response. Cognitive change acts upon the appraisal component of the model. It refers to the efforts made to revise the situation's emotional significance and meaning, in an attempt to influence one's emotions. That is, an individual may change how he/she thinks about the situation or about his/her capacity to manage situational demands. Finally, response modulation refers to the efforts made to change or influence one's behavioral, or physiological responses to the emotion-eliciting situation (Gross & Barrett, 2011; Gross & Jazaieri, 2014). In any given emotion-generative cycle, an individual moves in sequence through these processes and may engage in one or more ER strategies. It is important to note, however, that these five strategies may be adaptive or maladaptive – although entirely dependent on the individual, the emotion, and the situational context (Aldao & Nolen-Hoeksema, 2012).



**Figure 1.1.** The process model of emotion regulation. Adapted from "Emotion Regulation: Conceptual Foundations," by J. J. Gross and R. A. Thompson, 2007, *in Handbook of Emotion Regulation* (p. 10), J. J. Gross (Ed.), New York, NY, Guilford Press.

#### 1.3.2. Expressions of Emotional Regulation in Childhood

During infancy, ER emerges in its most basic form – progressing from a highly external process (i.e., caregiver) to an internal process (Thompson, 1994). During the first few months of life, infant reactivity is primarily innate, and comprised of reflexive emotional repertoires and discomfort related to physiological functioning. These repertoires may include responses to hunger, pain, fatigue, and cold temperatures. As the infant grows and develops, they begin to react to psychological distress (e.g., lack of contact with caregiver) and ER begins to shift from reflexive activity to neurophysiological modulation of arousal and behavior (Kopp, 1982). It is during this time that the infant depends largely on their caregivers for the regulation of their emotional state (i.e., is externally regulated; Crockenberg & Leerkes, 2006). Moreover, infants also begin to engage in simple self-regulatory behaviors, including avoidance, self-comforting

behaviors, non-nutritive suckling, and head-turning behaviors (Buss & Goldsmith, 1998; Crockenberg & Leerkes, 2004). By 3-4 months of age, the ability to use attentional strategies (i.e., attentional orienting, distraction, and gaze aversion) emerges – signifying a critical component in the regulation of emotional states as it demarks the onset of innate (self) regulation (Rothbart & Derryberry, 1981; Johnson et al., 1991). Simple regulatory strategies progress to reaching for objects, complex uses of gaze for attention regulation, and additional self-soothing behaviors (Kopp, 1989; Dawson et al., 1992). By the end of the first year, infants become goaldirected and their repertoire of regulation strategies are reflective of their growing cognitive capacities (Kopp, 1982).

During the toddler years, children develop a sense of agency and become increasingly aware of the causes of their distress. Mature forms of regulation emerge, including the use of discrete strategies (e.g., self-distraction, self-soothing, and goal-directed behaviors; Kopp, 1989; Mangelsdorf et al., 1995; Jahromi & Stifter, 2007). Moreover, constructive coping strategies emerge, reflective of increasing cognitive and language skills (Calkins & Johnson, 1998; Grolnick, Bridges, & Connell, 1996; Diener & Mangelsdorf, 1999; Cole et al., 2010). Gains in motor development that occur during the second year of life also enable toddlers to explore or withdraw from emotionally arousing stimuli and/or events. Toddlers also begin to use social referencing more frequently, guiding the appropriate usage of emotional expressions (Kopp, 1989).

As children progress into the preschool years, use of cognitive appraisals (with the help of a caregiver) begins to emerge, where children develop skills to reinterpret negative emotioneliciting stimuli and/or events (Stansbury & Sigman, 2000). Furthermore, the expression of negative affect significantly declines during this period, which is attributed to the child's improved ability to modulate emotional expressions during stressful contexts (Cole et al., 1994). Finally, during distressing situations, children employ a wide range of coping strategies to overcome these difficulties. Coping strategies may include distraction, help- and comfort-seeking behaviors, goal-directed strategies, and other forms of tension release (Diener & Mangelsdorf, 1999).

#### 1.3.3. Expressions of Emotion Regulation in Autism Spectrum Disorder

Difficulties in ER play a role in the emerging ASD phenotype. Although emotional dysregulation is not a core diagnostic feature of ASD (American Psychiatric Association, 2013), differences in ER have been identified in children with ASD, such as maladaptive emotional responses that can include self-injury, aggression, irritability, and temper tantrums (Lecavalier, 2006; Quek et al., 2012). Retrospective studies have indicated that although children with ASD do not appear less emotionally expressive than children with typical development (TD) or intellectual disability (Ricks & Wing, 1975), they do show higher levels of negative emotions and lower levels of positive emotions (Capps et al., 1993). Similarly, compared to children with TD or intellectual disability, children with ASD demonstrated deficits in the sharing of emotional experience, such as reduced positive affect and coordination of gaze with emotion expression to share experiences (Yirmiya et al., 1989; Kasari et al., 1990, 1993). Furthermore, prospective studies of children later diagnosed with ASD have noted that reduced social-emotional reciprocity, in conjunction with delays in verbal and non-verbal communication, contributed to increased negative affect, reduced positive affect, and lower levels of regulation (Gomez & Baird, 2005; Zwaigenbaum et al. 2005; Bryson et al., 2007; Clifford et al., 2007; Watson et al., 2007; Garon et al. 2009, 2016). These atypicalities are apparent in the first years of life, with approximately 30% of parents reporting ER difficulties by their child's first birthday (De Giacomo & Fombonne 1998; Sacrey et al., 2015).

In line with these findings, examination of ER in school-aged children with ASD also highlight the increased prevalence of ER difficulties compared to TD peers. That is, children with ASD and intellectual disability have a higher prevalence of emotional disorders than children with intellectual disability but without ASD (Totsika et al., 2011). In contrast, intellectually-able children with ASD show a similar range of expressiveness as TD peers, yet they also display less positive affect (Capps et al., 1993) and have higher levels of emotional and conduct problems than their TD peers (Totsika et al., 2011). Furthermore, research has demonstrated that children with ASD use less adaptive self-regulatory coping strategies (e.g., crying, avoidance) when exposed to mildly frustrating situations compared to their TD peers (Jahromi et al., 2012; Konstantarias & Stewart, 2006). Similarly, Zantinge and colleagues (2017) found that children with ASD demonstrated similar physiological responses to frustrating stimuli; however, their behavioral coping strategies were less constructive (i.e., did not use goaldirected behavior, social support seeking, or orienting to caregiver) than those used by the TD peers.

#### 1.4. Social Communication

Social communication encompasses the knowledge, skills, and behavior necessary to engage in social interactions (Hwa-Froelich, 2015), including understanding and use of verbal and non-verbal communication, as well as the flexible implementation of these skills during reciprocal interactions with others (Goldstein & Morgan, 2002; Muller, 2000; Muma, 1991; Gallagher, 1991; Ninio & Snow, 1996). Dynamic reciprocal social interactions require a child to engage in social exchanges, have social awareness, engage in social information processing and cognition, and have the capacity for social interaction (Constantino, 2002).

In the first few years of life, the ability to engage in social interactions develops as infants learn to respond to their social partners and regulate their behavior in response. One of the earliest forms of social communication that develops in infancy is joint attention (Bakeman & Adamson, 1984; Baldwin, 1995; Bruner, 1975; Tomasello, 1995). Joint attention is an umbrella term that refers to a set of behavior including eye gaze, pointing, and showing, that are used to incorporate objects, people, and events into a communicative exchange (Carpenter et al., 1998; Charman, 2003) and allows infants to coordinate their attention with their social partner in the context of social interactions. Measures of joint attention are widely used in research on the development of both typical (Tomasello & Farrar, 1986; Mundy, Sigman, & Kasari, 1990; Ulvund & Smith, 1996; Morales, Mundy, & Rojas, 1998) and atypical populations (Wetherby & Prutting, 1984; Loveland & Landry, 1986; Mundy, Sigman, Ungerer, & Sherman, 1986; Sigman & Ruskin, 1999; Mundy & Neal, 2001). Gaining competence in this fundamental skill undergirds later language development and ER.

Joint attention deficits are precursors for later social and language skills in children with ASD (Charman, 2003; Poon et al., 2012). For example, Travis and colleagues (2001) reported that initiating joint attention was associated with peer engagement and prosocial behavior in 8- to 15-year-old children with ASD. Furthermore, Sigman and Ruskin (1999) reported that measures of initiating joint attention at 6 to 8 years in children with ASD was associated with social engagement at 12 to 14 years of age, even when controlling for cognitive and language development. Measures of responding to joint attention were not related with social

development, but were related to language acquisition. This is in line with reports that deficits in joint attention are thought to contribute to poor language outcomes in ASD (Anderson et al., 2007; Norrelgen et al., 2014, Tager-Flusberg et al., 2005), with over 60% of children with ASD also having co-occurring language disorders (Levy et al., 2010).

#### 1.5. Association between Emotion Regulation and Social Communication

Young children rapidly develop social and emotional competencies that allow them to navigate increasingly complex social interactions, including establishing and maintaining relationships with others - skills that are important contributors to emotional well-being and mental health in adulthood (Frijda, 1986; Ekman, 1992; Saarni et al., 2006). Social communication and ER are enmeshed and impact how a child makes social connections and responds to social partners (Frijda, 1986; Ekman, 1992; Saarni et al., 2006; Darwin et al. 1998; Shariff & Tracy, 2011; Cole & Moore, 2015). During development, the expression of emotions is regarded as the first social communicative action of infants. Emotional expressiveness in infancy evokes responses from caregivers, inviting them to share emotional states and engage in socialemotional reciprocity. By the second year, intentional communication skills develop, allowing toddlers to engage in complex behavior to share experiences with others (Mundy, 1995). The degree of scaffolding and responsiveness by caregivers helps to regulate emotions and impacts subsequent social-emotional development (Carter et al., 2004; Feldman, 2007).

Social communication and ER are fundamental skills, thus it is essential to investigate how the relationships between ER and social communication are impacted in young children with ASD. This relationship has been explored in older children with ASD. For example, maladaptive ER has been associated with mental health in children with ASD, including attention problems, aggression, depression, and anxiety (Patel et al., 2016,; Pouw et al., 2013; Rieffe et al., 2011). Investigating ER in infants and children at an increased likelihood for ASD is imperative to further our understanding of their relationship and how they impact later ASD emergence.

# 1.6. Theoretical Frameworks Guiding the Relationship Between Emotion Regulation and Social-Communication

Joint attention and ER are important factors in the development of later skills, yet the developmental pathways underlying these processes are not fully elucidated. To further our understanding of the relationships between early ER and joint attention, three theoretical frameworks will be reviewed: (1) Domain-Relevant Framework, (2) Temperament Framework and, (3) Parallel and Distributed Processing Model (PDPM) of Joint Attention. These theoretical frameworks – ranging from general to more specific concepts – have guided the present dissertation work.

#### 1.6.1. Neoconstructivist Approach: Domain-Relevant Framework

It is a truism that evolution and ontogenetic development involves contributions from both genes and the environment. Yet, the complex interaction and degree to which genes and the environment contribute to developmental outcomes has been largely debated among theorists. An empiricist would argue that the infant brain is a 'blank slate' upon which environmental experience imprints itself via *domain-general* learning mechanisms (i.e., infant development is largely dependent on interactions with the environment; Elman et al., 1996; Kirkham, Slemmer & Johnson, 2002; Lany & Saffran, 2013). Nativists, on the other hand, propose that the infant brain comes equipped with innate, built-in *domain-specific* knowledge systems or modules upon which more knowledge is built through learning (Baillargeon & Carey, 2012; Carey, 2011; Spelke & Kinzler, 2007; van der Lely, 2005; van der Lely & Pinker, 2014). Are these two extremes the only frameworks for theorizing about ontogenesis and subsequent developmental outcomes (such as the development of emotional and social systems)? Karmiloff-Smith argues for a middle ground with her neoconstructivist approach – referred to as the *Domain-Relevant* framework – for understanding functional specialization in the developing brain.

The *Domain-relevant* framework argues that the infant brain is neither an experiential learning device nor one that contains numerous domain-specific knowledge systems (Karmiloff-Smith, 2009). Rather, it proposes that the developing brain begins with a number of basic-level biases that are relevant to, but not initially specific for, processing different types of environmental input. These *Domain-relevant* biases become *domain-specific* over time via extensive environmental experience and neuronal competition (Elman et al., 1996; Johnson, 2001; Karmiloff-Smith, 1992, 1998, 2009, 2012, 2015; Westerman et al., 2007, 2010). In other words, functional specialization of brain regions, neural networks, and systems emerge gradually

as a result of developmental processes, as opposed to a domain-specific start state (De Souza et al., 2017). This perspective of the brain as a plastic and self-structuring system – vulnerable to environmental influence at cellular, neural, cognitive, and behavioral levels over time – provides a framework for the development of emotional, attentional, and social systems underlying the present thesis work. For example, it can be speculated that the infant brain displays more widespread neural activity in response to, say, early emotional or joint attention-eliciting stimuli. Following extensive experience over infancy, neural processing of emotion and joint attention (in response to stimuli) becomes increasingly localized and specialized. That is, what was *Domain-Relevant* to processing of emotion and joint attention becomes *domain-specific* as a result of repeated processing, interaction, and competition of these inputs. This form of gradual developmental localization and specialization of neural function throughout infancy likely underlies the development, interaction, and flow of information between these two systems, and may have effects on later-developing neural systems and behavior (such as refinement of the social brain and language abilities).

*Implications for Dissertation.* The *Domain-Relevant* framework likely guides the ontogenesis and functional specialization of emotional and socio-communicative systems, where *Domain-Relevant* biases kick-start development, followed by neuronal competition and environmental experience to refine the neural networks and connection between these systems. Using this framework, I would predict that ER would impact the development of social communication and vice versa, indicating a bi-directional relationship.

#### 1.6.2. Temperament Framework

Beginning in infancy, children exhibit observable differences in how they respond to and interact with their environment. Rothbart and Bates (2006) refer to these individual differences as temperament. Temperament refers to "constitutionally based individual differences in *reactivity* and *self-regulation* in the domains of emotion, activity, and attention" (Rothbart & Derryberry, 1981). The term 'constitutional' implies that temperament is biologically based and influenced by genes, experience, and maturation over developmental time. Temperamental *reactivity* is defined as responses to changes in the internal and external environment and may encompass a wide range of reactions, such as negative affect, orienting, motor activity, cardiac reactivity, etc. Thus, *reactivity* is measured with respect to the duration, speed, and intensity of

the emotional, motor, and orienting reactions (Rothbart & Derryberry, 1981; Rothbart & Sheese, 2007). Temperamental *self-regulation*, on the other hand, refers to the processes that modulate *reactivity*.

A child's temperamental *reactive* tendencies to experience and express emotions, as well as his/her responsivity to environmental stimuli/events, can be observed early in infancy. In fact, each of these reactive tendencies (i.e., emotional, motor, and orienting responses) contains selfregulatory aspects. For example, the emotion of fear is a reactive temperamental system that may potentiate attack, withdrawal, or inhibit behavior. Positive affect, on the other hand, may potentiate rapid and energetic approach behavior (Rothbart, 2011). Early attentional orienting is another *reactive* temperamental system. It exerts regulatory effects on an infant's emotional expression when a social partner presents distracting stimuli to modulate distress, for example (Harman, Rothbart, & Posner, 1997). These examples highlight that individual differences in the intensity and temporal patterns of the emotional responses are not only assessed when examining temperamental *reactivity*, but differences in the organization and functioning of the emotionprocessing networks of these responses ought to also be considered (Rothbart & Sheese, 2007). In fact, Derryberry and Rothbart (1997) propose two emotion-processing systems that represent the self-regulatory and reactive components of temperament: the defense system and the approach system. First, the defense system is comprised of a series of brain networks (e.g., lateral and central amygdala) that serve the goal of avoiding harm by activating organized responses to immediate and long-term threats/stimuli. In other words, activation of the defense system may evoke fear, anxiety, and defensive anger responses (i.e., a "fight" response), or elicit withdrawal behavioral tendencies to avoid potentially threatening situations (i.e., "flight" response). Aspects of the defense system, including fearful behavioral inhibition, develop late in the first year of life and show longitudinal stability across childhood and adolescence (Kagan, 1998). The second emotion-processing system is referred to as the approach system, which serves the goal of resource acquisition by promoting behavioral tendencies to seek out and approach rewards. In other words, activation of the approach system evokes positive emotional states (i.e., surgency/extraversion), such as joy or eager anticipation, in response to reward acquisition. Comprised of the amygdala, ventral tegmental area, and nucleus accumbens, aspects of the approach system – including positive affect and approach to novel objects – develops by 3 months and exhibits stability from 3-9 months of age (Rothbart, Derryberry, & Hershey, 2000).

The development of temperamental self-regulation is largely dependent on the development of attention, including the control of orienting and the development of executive attention (Rueda et al., 2004). Specifically, executive attention provides the neural substrate for developing effortful control. Effortful control is defined as the ability to voluntarily regulate attention and behavior by inhibiting a dominant response in order to perform a subdominant response, detecting errors, and engaging in planning (Rothbart & Rueda, 2005). In the early months of life, an infant's primary concern is the regulation of state, particularly the regulation of distress. The modulation of distress occurs via selective orienting of attention (i.e., early form of joint attention), where the caregiver utilizes soothing techniques that include distracting the infant by bringing their attention to other stimuli (Halsted, 1991). These techniques provide a major basis for later self-regulation in both orienting and executive attention. Beyond infancy and into the toddler and preschool years, children become skilled at making increasingly difficult adjustments in their thoughts and behavior via effortful control. That is, as language and improved impulse control become available to the child, improvements in executive attention and self-regulation follow (Kopp, 1992). It is proposed that the development of the executive attention system underlies effortful control, whereby children attain greater control and flexibility of behavioral response, including the ability to select responses in a conflict situation (Rothbart & Sheesh, 2007; Rothbart et al., 2006). Effortful control, therefore, is not only important for the regulation of emotions and behavior, but is also critical to socialization and the development of social competence. In fact, effortful control has been positively linked to empathy, guilt, aggression, and conscience (i.e., issues central to socialization; Posner & Rothbart, 2000). Overall, the study of temperamental individual differences highlight the importance of considering the underlying constructs of executive attention and effortful control in shaping a child's emotional, cognitive, and social development.

*Implications for Dissertation.* The *Temperament* framework is well suited given that the construct of temperament itself is comprised of ER and attentional networks (that is, ER influences attention, and attention influences ER). Using this framework, I would predict that ER and social communication are directly related and work together to impact the emergence of ASD symptoms.

#### 1.6.3. Parallel and Distributed Processing Model (PDPM) of Joint Attention

Mundy and colleagues proposed that an understanding of the nature of joint attention is central to the development of cognitive, attentional, emotional, and social systems. Defined as an information processing system, joint attention reflects the degree to which an infant coordinates his/her attention with that of a social partner in order to share a common experience (Bruner, 1975; Mundy et al., 2007; Mundy & Newell, 2007). Joint attention involves a three-way deployment of attention between the infant, a social partner, and an object/event, as well as triadic information processing. That is, joint attention requires an infant to attend to and process (1) information regarding an object/event, (2) information regarding the social partner's attention and behavior in relation to the object/event (e.g., eye gaze), and (3) self-referenced information related to one's attention to and experience of the object/event and the situation (Mundy et al., 2010). This self-referenced information processing, in particular, involves subjective processing and integration of both interoceptive (i.e., physiological information including heart rate, autonomic arousal, and emotional states) and proprioceptive (i.e., sensitivity to the location, position, and movement of the body) information from the infant's own body (Northoff et al., 2006; Zahavi, 2003). Continual practice of this joint processing of attentional information from oneself and others throughout infancy enables infants to socially coordinate their attention with social partners, which is fundamental to social learning and later social competence (Baldwin, 1995; Bruner, 1975; Striano et al., 2006 a, b).

Within the first few months of life, two forms of joint attention emerge. Responding to Joint Attention (RJA) reflects an infant's ability to follow the direction of gaze, gestures, or head posture of a social partner, resulting in information processing of others signals and a shared social point of visual reference. RJA is thought to be associated with the posterior cortical attention network (i.e., neural networks of the parietal and temporal cortices) and actively involved in controlling orienting behavior, as well as the development of cognitive representations of the world based on information obtained from external senses (Dosenbach et al., 2007; Cavana & Trimble, 2006; Fuster, 2006; Dosenbach et al., 2007; Mundy & Newell, 2007; Mundy et al., 2009; Posner & Rothbart, 2007). Initiating Joint Attention (IJA), on the other hand, refers to an infant's ability to spontaneously initiate a shared point of reference via alternating gaze between objects/events and social partners, or by the use of gestures. Research has indicated that IJA is associated with the later-developing anterior cortical attention network (i.e., anterior cingulate, anterior prefrontal cortex, orbital frontal cortex, basal ganglia, and medial superior frontal cortex including the frontal eye fields) and is implicated in the cognitive processing, representation, and regulation of volitional goal-directed behavior (Dawson et al. 2004; Fuster, 2006; Dosenbach et al., 2007; Posner & Rothbart, 2007; Mundy et al., 2009). While it is evident that both RJA and IJA interact and integrate during early development, it is important to bear in mind that these forms of joint attention also dissociate in development (given key differences in function and developmental timing of the posterior and anterior attention networks; Mundy et al., 2000, 2007). That is, while stable individual differences in both RJA and IJA are established by 8 to 9 months of age, there is a dissociation in developmental onset of these behavior (where RJA emerges in the first 3 months of life and IJA develops between 4-5 months of age; Venezia et al., 2004; Striano & Reid, 2006; Mundy et al., 2007; Mundy & Vaughan van Hecke, 2008). Moreover, the parallel integration of internal information about an infant's own visual attention with information regarding visual attention of a social partner across a distributed cortical network is the defining feature of joint attention (Mundy & Newell, 2007). This distributed and parallel attention system has implications for the development and maturation of emotional, attentional, and social systems underlying the current dissertation.

With respect to the development of the emotional system, early joint attention also plays a critical role in ER (Rothbart & Posner, 2006). That is, to move attention from one visual stimulus to another, an infant must learn to first *disengage* from current stimuli, then *shift* and *engage* attention at the new location. The ability to disengage typically develops by 4-6 months of age, allowing infants to redirect attention from arousing events and thereby regulate their emotional state. For example, if an infant has difficulties disengaging from negative stimuli, this may present as negative affect and a behavior response. In contrast, typical disengagement from negative stimuli would likely contribute to the development of positive or neutral affect and its associated behavioral response. In the second half of the first year, executive control over attention emerges and ER improves (Colombo & Cheatham, 2006). This may reflect a dynamic and concurrent relationship between joint attention and ER development, whereby these systems interact in reciprocal cause-and-effect relationship alongside experience.

With these important features in mind, Mundy and colleagues (2009, 2010) proposed a novel model of joint attention, referred to as the *Parallel and Distributed Processing Model* (PDPM) of Joint Attention. The PDPM is believed to lay the foundation for the development of

social cognition and social competence. According to this model, joint attention involves the parallel processing of self-referenced information (interoception *and* proprioception), information regarding a social partner's attention, and information regarding a commonly referenced object/event. An infant's continued experience and practice with the processing of self-, social partner-, and object/event-referenced information in infancy becomes affected by, and has an effect on the development of a distal distributed cortical system. This cortical system involves the posterior cortical attention system (related to the behavior of social partners) and the anterior cortical attention system (related to goal-directed behavior and/or attention of self). As development progresses and an infant is provided with further experience, the repeated and simultaneous activation of both the anterior and posterior cortical attention systems increases association between the two cortical systems (Hebb, 1949). The result is a parallel and distributed neural network that serves a social-executive function: it allows for seamless coordination of attention during social interactions. Thus, an infant's capacity to integrate and share overt aspects of attention lays the foundation for the ability to share covert aspects of attention and social cognition.

*Implications for Dissertation.* The *PDPM* framework allows for the examination of the effects of early ER on RJA and IJA independently, rather than a composite measure of joint attention, and their impact on subsequent ASD emergence. Using this framework, I would predict that ER will differentially influence IJA and RJA (due to different cortical organizations), which in turn may have differential impacts on the emergence of ASD symptoms.

#### 1.7. Current Dissertation

The present dissertation consists of three studies designed to identify the developmental relationships between ER and joint attention in infancy and how they influence the emergence of ASD. The overarching research question is: *What are the developmental relationships between early ER and social communication in ASD*? The overarching framework is depicted in Figure 1.2.

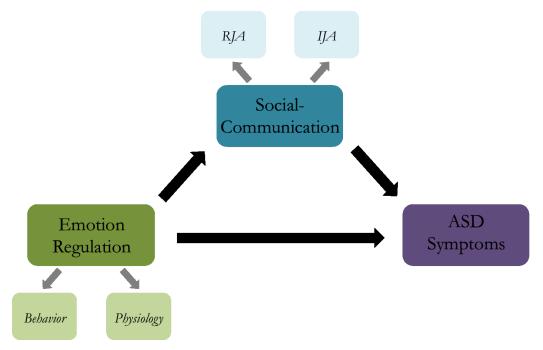


Figure 1.2. Overarching research framework.

#### 1.7.1 Dissertation Research Questions and Hypotheses

#### Chapter Two.

<u>Research Question</u>: What are the relationships between early ER and social communication development in childhood?

*Hypothesis:* There is a bidirectional relationship between ER and social communication in early childhood, with each construct influencing the other throughout development.

#### Chapter Three.

**Research Question:** What is the relationship between behavioral affect and physiological arousal during an ER task in a cohort of infants who are at an increased likelihood (IL) and low likelihood (LL) of ASD?

*Hypothesis 1*: The relationship between behavioral affect and physiological arousal will be differentially impacted in LL versus IL infants.

*Hypothesis 2:* Affective and physiological congruence in the IL group will predict later ASD symptoms, but not the LL group.

### Chapter Four.

<u>Research Question</u>: Do variations in behavioral and physiological indices of ER in infancy predict subsequent joint attention in IL and LL infants?

*Hypothesis 1:* Infants in the IL group will display atypical ER and lower rates of joint attention compared to LL infants.

*Hypothesis 2*: ER and joint attention will predict subsequent ASD symptoms at 24 months.

*Hypothesis 3*: Joint attention will mediate the relationship between ER and later ASD symptoms.

## CHAPTER 2: The Bidirectional Relationship Between Emotion Regulation and Social Communication in Childhood: A Systematic Review

The construct of emotion regulation (ER) has assumed a central role in the developmental psychology literature. Although there is debate regarding the conceptualization and precise definition of ER, theorists generally agree that ER involves the dynamic interplay of affective, behavioral, and cognitive processes used to regulate emotional reactions to environmental stimuli (Calkins, 1994; Calkins & Hill, 2007; Cole et al., 2004; Eisenberg et al., 1995, 1996; Kopp, 1982; Thompson, 1994). ER begins in the first year of life, whereby a child uses behaviors and skills to modulate, inhibit, or enhance their emotional responses – skills that become increasingly complex throughout development. That is, ER may involve intrinsic or extrinsic strategies (i.e., learned versus assistance from others), be conscious or unconscious, as well as automatic or controlled (Calkins & Hill, 2007; Cole et al., 1994; Gross & Thompson, 2007).

ER contributes to the development of social communication – the knowledge, skills, and behavior necessary to engage in reciprocal social interactions (Hwa-Froelich, 2015) - which has implications for social competence, decision-making, and attention (Calkins, 1994; Calkins & Hill, 2007; Cole et al., 2004; Eisenberg et al., 1995; Kopp, 1982; Thompson, 1994). In fact, ER has strong origins in early social interactions, particularly between infants and their caregivers (Kopp, 1989; Thompson, 1988). That is, the expression and regulation of emotion is regarded as the first social communicative action of infants. Through the use of gestures, eye gaze, and vocalizations, infants communicate their emotional states and needs to their caregivers (Bates et al., 1979; Trevarthen & Aitken, 2001; Volterra et al., 2005). As these skills become integrated, young children begin to engage in sustained reciprocal interactions, while sequentially regulating their emotional states (Iverson & Thal, 1998). It is this interaction between early ER and social communication that allows young children to navigate increasingly complex social interactions. In other words, ER and social communication are dynamically linked and interact to help young children process information and engage in emotion-regulatory behavior (Frijda, 1986; Ekman, 1992; Saarni et al., 1998; Bell & Wolfe, 2004; Darwin et al. 2006; Shariff & Tracy, 2011; Cole & Moore, 2015).

Understanding the developmental relationships between ER and social communication are important, as these skills form the foundation for school success, building lasting

relationships with family and friends, and navigating social environments (Pontoppidan, 2017). As a child learns to differentiate and regulate his/her emotions, he/she also develops the capacity to recognize the emotional states of others to determine how to engage in social situations. For example, if a child is unable to regulate his/her anger towards a peer, the subsequent interaction may be fragmented – resulting in disrupted play or escalating conflict. Likewise, if a child is unable to understand the emotional states of others, he/she may also have difficulties in the regulation of his/her own emotions, resulting in disturbances in interpersonal interactions (Fabes & Eisenberg, 1992; Slomkowski & Dunn, 1996). Therefore, it is critical to examine ER and social communication in tandem to better understand how these processes affect functional outcomes. It is clear that these processes are fundamentally intertwined; however, ER and social communication are often studied separately. Examining the bidirectional relationship will help us better understand both normative and non-normative trajectories.

The purpose of this systematic review is to summarize research that has examined the temporal and bi-directional relationship(s) between ER and social communication in childhood. Specifically, the goals are to: (1) identify the constructs and measures used to assess childhood ER and social communication, (2) describe the relationship between early ER (i.e.,  $\leq 2$  years of age) and subsequent social communication, (2) describe the relationship between early social communication (i.e.,  $\leq 2$  years of age) and subsequent ER, and (3) calculate effect size estimates to evaluate the strength and patterns of these temporal relationships. We then discuss the implications of the bi-directional relationship between ER and social communication and how these developmental relationships are important for later socio-emotional functioning and child mental health.

#### **METHODS**

#### Search Strategy

In accordance with the guidelines of Preferred Reporting of Items for Systematic Reviews and Meta-Analyses (PRISMA; Moher et al., 2009), a systematic search in Medline Ovid, PsychINFO, Scopus, and CINAHL was performed from database inception until March 20, 2022. Search strategy and terms were refined in collaboration with a University of Alberta health sciences librarian. The search strategy included a combination of subject headings and keywords to combine concepts of (1) emotion, (2) emotion regulation, (3) social communication, (4) infant, and (5) longitudinal study design. The Scottish Intercollegiate Guidelines Network (SIGN) search filter for Observational Studies was also applied, which contained index terms related to observational studies and methodological descriptions of good experimental design (https://www.sign.ac.uk/search-filters.html). The full search strategy is available in Appendix 1. Search results were imported into Covidence (covidence.org) for review.

#### Screening

To be included for review, articles were required to meet the following inclusion criteria: (1) examine the relationship between ER and social communication, (2) include distinct measures or tasks of ER and social communication, (3) include at least one time point in infancy ( $\leq 2$  years of age), (4) include a sample of typically developing children, and (5) possess a longitudinal study design. Articles were excluded if they were: (1) not data-based (e.g., case reports, reviews, books, commentary), (2) intervention studies, (3) examined children with pediatric disorders but did not include a typically developing comparison group, or (4) did not include direct measures of ER (e.g., attachment, shyness, etc.) or social communication (e.g., reticence, wariness, etc.).

Initial literature search resulted in 3,672 articles identified (1,034 from Medline, 875 from PsychINFO, 967 from Scopus, and 796 from CINAHL). After removal of duplicates, the titles and abstracts of 2,594 articles were independently screened by two reviewers (SR and LRS) using the inclusion and exclusion criteria. Any disagreement between the two reviewers was resolved by a third reviewer (LZ). Resultant from this screen, 2,364 articles were further excluded, with a total of 230 unique articles eligible for full-text review. The first author (SR) completed a full-text review of the 230 articles, with 207 articles being further excluded. Reasons for exclusion included focus on only one of the two essential constructs (i.e., ER or social communication, but not both), study design (i.e., not longitudinal), outside desired age range, patient populations with no typically developing comparison group, and non-data-based studies. Despite thorough search with a health sciences librarian and attempted communication with article authors, six articles could not be obtained (Wilde, 1983; Adrien et al., 1992; D'Entremont, 1996; Lavelli, 1996; Korntheuer, Lissmann, & Lohaus, 2010; Lavelli & Barachetti, 2010). A total of 23 articles met criteria and were included in the present review. Figure 1 details the search strategy and reasons for exclusion, adopted from PRISMA.

#### **Data Extraction**

SR and LRS developed a structured extraction sheet to collect relevant information and ensure comprehensiveness in data extraction. Study demographics were recorded and compiled, including year of publication, study objective, sample size, participant age range and sex, and comparison groups (if applicable). Articles were also reviewed for methodology and results, including ER and social communication measures, mode of assessment (e.g., observational, questionnaire, etc.), constructs/domains of ER and social communication assessed, temporal direction of the relationship between ER and social communication, and study results and effect sizes. Where unavailable, effect sizes (Cohen's *d*) were calculated from eligible studies using methodology (i.e., from the means and standard deviations) drawn from Rosnow & Rosenthal (1996), Rosnow et al. (2000), and Thalheimer & Cook (2002). Classification of Cohen's *d* was 0.20 - 0.49 = small effect, 0.50 - 0.79 = moderate effect, and  $\ge 0.80 =$  large effect (Cohen, 1988). For the purposes of this review, only findings pertaining to ER and social communication were extracted. Some of the included studies investigated a broader range of developmental constructs, but these will not be reported.

#### **RESULTS**

The systematic review examining the developmental relationships between ER and social communication resulted in the inclusion of 23 articles. The results will first provide a brief overview of included articles, followed by a narrative synthesis of the bi-directional relationships between ER and social communication. That is, results were organized into two broad sections: (1) relationship between early ER and subsequent social communication and (2) relationship between early social communication and subsequent ER. Within each section, studies were further categorized by type of assessment: (a) questionnaire measures and (b) observational measures. Assessments utilized were briefly described, followed by key findings and results related to the ER and social communication relationship. Among studies that examined both directional relationships (i.e., ER to social communication and social communication to ER), study findings were included in both sections. Descriptive details and study demographics are presented in Table 2.1; Table 2.2 outlines the methodology used in the studies; Table 2.3 provides descriptive details of questionnaires and observational measures; and Table 2.4 outlines the results of the studies.

#### **Overview of Included Articles**

There was a total of 23 articles included in the present systematic review, with the earliest article meeting screening criteria published in 1982 and the most recent published in 2018. The majority of articles originated from the United States (n=16), with the remainder from the Netherlands (n=1), Sweden (n=1), France (n=1), Italy (n=1), Canada (n=1), Korea (n=1), and Taiwan (n=1). Of the 23 articles, 21 included a single typical developing sample, whereas two articles included comparison groups – preterm infants (Hwang et al., 2009) and infant siblings of autism spectrum disorder (Cassel, 2008). For the purpose of this review, only findings pertaining to the typically developing group were examined. Study sample sizes ranged from 26 (Parlade et al., 2009) to 6,850 participants (Rispoli et al., 2013). Moreover, age of participants ranged from as early as one month of age (Bowers et al., 2018) to eight years of age (Bohlin et al., 2005; Bowers et al., 2018).

Overall, 19 studies assessed the relationship between ER and subsequent social communication; one examined the relationship between social communication and subsequent ER; and three examined bi-directional relationships. There were a total of 17 different ER measures and 21 social communication measures used across the 23 studies, with many studies employing more than one measure. Across the ER measures, 53% utilized at least one parent or teacher reported measure and 47% included at least one naturalistic observation. Among the social communication measures, 57% included at least one parent or teacher reported measure and 43% utilized at least one naturalistic observation measure and 43% utilized at least one naturalistic observation measure measure.

## 1. Relationship Between Early ER and Subsequent Social Communication-Related Constructs

#### A. Studies using Questionnaire Measures of ER

The majority of studies that examined the relationship between early ER and subsequent social communication used questionnaire measures of ER, specifically temperament-related questionnaires. When examining the directional relationship between ER and subsequent social

communication, there were 15 studies that used temperament questionnaires to index early ER, as detailed in Tables 2.3 and 2.4.

#### Studies using Rothbart, Goldsmith Questionnaire Measures of ER

Rothbart, Goldsmith, and colleagues developed a series of questionnaires designed to measure early childhood temperament, including the Infant Behavior Questionnaire (IBQ), the Toddler Behavior Questionnaire (TBAQ), the Early Child Behavior Questionnaire (ECBQ), and the Children Behavior Questionnaire (CBQ), as described in Table 3. Ten studies used these questionnaires (Thompson & Lamb, 1982; Henderson et al., 2001; Karass, 2002; Albers et al., 2007; Van Hecke et al., 2007; Markova, 2008; Penela et al., 2012; Salley et al., 2013; Penela et al., 2015; Aureli et al., 2017).

Albers and colleagues (2007) examined the relationships between the IBQ-R and social play and interactions to investigate if *negative emotionality* in 3-month-olds was associated with the quality of social interaction with caregivers of childcare centers at 6 month of age. Infant negative emotionality was rated by mothers at 3 months of age using the IBQ-R, and infantcaregiver behavioral interactions were examined during caregiving episodes (e.g., changing diapers, feeding, putting to bed) at 6 months of age for ratings of infant responsiveness to caregiver (e.g. responding to bids) and *involvement* (e.g., initiating contact). Interestingly, infant *negative emotionality* was not associated with infant *responsiveness* (d=0.06) nor *involvement* (d=0.06). The lack of relationship between temperament and social communication did not appear to stem from the caregiver. Similarly, a longitudinal examination of the relationship between infant temperament at 4 months of age was also not associated with mother-child interactions at 6 months of age (Aureli et al., 2017). Aureli et al. (2017) examined infant temperament using the *cuddliness*, soothability, perceptual sensitivity, low intensity pleasure, and duration of orienting scales of the IBQ-R (Italian version) at 4 months and mother-child communicative interactions during face-to-face/dyadic and object/triadic play at 6 months of age. There were no significant relationships between any of the temperament subscales, with the exception of *soothability*. Soothability at 4 months was positively correlated with symmetrical co-regulation (i.e., mother and infant mutually engage in activity) at 6 months during triadic interactions, revealing a moderate relationship (d=0.36).

Although maternal caregiving was not directly related to social communication, maternal caregiving appeared to play a mediating role between infant fear and social engagement and global aggression in the Penela et al. (2012) study. Infant temperamental fear was measured at 9 months of age using the IBQ, and social engagement was measured during free play with an unfamiliar, same-sex peer at 2 years of age. Overall, levels of *fear* at 9 months of age were associated with social engagement at age 2, with high levels of fear predicting lower levels of social engagement with an unfamiliar peer, but only when moderated by low maternal caregiving behavior (d=0.20). Similarly, low levels of *fear* were associated with more *aggression*, only when moderated by low maternal care (d=0.17). These findings were corroborated by Henderson et al. (2001), who examined the relationship between *negative reactivity* and *sociability* during peer play. Maternal reports of *negative reactivity* at 9 months of age were measured using *fear* and *distress to limitations* subscales on the IBQ and compared to a *sociability* aggregate score at 4 years of age, computed from social behavior during unstructured free play with unfamiliar peers and maternal report on the Colorado Child Temperament Inventory (CCTI) sociability scale. With a moderate effect size (d=0.35), the authors found that *negative reactivity* at 9 months of age did not predict *sociability* at age 4, although the relation was in the expected negative direction.

The lack of direct relationship between infant temperament and later sociability was further explored by Markova (2008). Broad temperament dimensions of *negative affectivity*, *extraversion*, and *regulation* were measured at 6 months using the IBQ-R. Infant social competence with peers was assessed at 7, 9, and 12 months and was operationalized as interaction skills (*gaze, facial expressions, vocalizations, initiations, responses to initiations, coordinated attention, communicative gestures, touch, reach,* and *play*) and *dyadic preferences* for peers (i.e., infant standing in peer group). Overall, there was no significant effect of any temperament dimension on the interaction skills of *gaze, positive affect, initiations* and *responses,* and *play*. Moreover, there were no effects of early temperament on *communicative gestures*; however, there was a significant interaction between *extraversion* and age (d=0.52), with infants rated as *extraverted* using more *communicative gestures* at 12 months. Interestingly, Markova (2008) did find that *negative affectivity* was significantly related to one aspect of sociality – *coordinated attention*. Infants rated high on *negative affectivity* engaged in less *coordinated attention* with peers overall (d=0.47). An interaction with age was also significant (d=0.01), where negative affectivity on coordinated attention was stronger at 12 months. With respect to the effect of early temperament on subsequent dyadic preferences, temperament was able to predict infants' standing in the peer group. That is, at 9 months, infants rated high on *negative affectivity* were more likely to be excluded (d=0.42), whereas *extraverted* infants were more likely to be included in a preferred dyad at 12 months (d=0.06). Despite the significant relationship between *negative affectivity* and later coordinated attention reported by Markova (2008), this finding was uncorroborated by Karass (2002). Karass (2002) assessed the relation between temperament and subsequent joint attention and found no relationship. Temperament composites of *positive emotionality* (*duration of orienting, smiling and laughter, soothability*) and negative emotionality (activity level, distress to limitations, distress to novelty) were measured at 4, 8, 12, and 16 months using the IBQ. Duration of time spent in joint attention during mother-infant free play was measured at 12 and 16 months. Positive emotionality at 4, 8, and 12 months was unrelated to *joint attention* at 12 months (d's=0.30-0.32) and 16 months (d's=0-0.08). Similarly, negative emotionality at 4, 8, and 12 months was unrelated to joint attention at 12 months (d's=0-0.24) and 16 months (d's=0.02-0.22). With respect to individual temperament dimensions, there were no significant correlations between early temperament and later joint attention.

The above studies assessed temperament prior to 12 months of age and did not find direct relationships between constructs on the IBQ and IBQ-R and later sociability (with the exception of dyadic preferences for peers). In contrast, Thompson and Lamb (1982) assessed measures of temperament at 12.5 months of age and found a direct relationship with *sociability* at 19.5 months of age. Measures of temperament, including *activity level, distress to limitations, fear, duration of orienting, smiling and laughter,* and *soothability* were collected at 12.5 months of age using the IBQ and compared to measures of *sociability*, as assessed by the Stranger Sociability procedure (adopted from Stevenson & Lamb, 1979) at 19.5 months of age. *Sociability* was negatively associated with *activity level (d=0.75), distress to limitations (d=0.93),* and *fear (d=0.63),* indicating a direct relationship in which temperament plays an important role in the development of stranger *sociability*. Assessment of temperament after age 2 was similarly associated with later sociability during free play with unfamiliar peers (Penela et al., 2015). Temperament was assessed through a latent construct for *behavioral inhibition,* derived from behavioral responses to unfamiliar stimuli and the TBAQ subscale, *social fearfulness,* at ages 2

and 3. As well, ER strategies (i.e., *active regulation, passive toleration*) were used to derive an engaged *emotion regulation* latent variable at age 5 during participation in a disappointing context task. Sociability was measured during unstructured free-play with unfamiliar peers at age 7, with positive social initiations, responses to initiations, and level of play coded to derive a *social competence* latent variable. *Behavioral inhibition* at 2-3 years of age was not directly related to *social competence* (d=0.01) at 7 years of age, but *emotion regulation* strategies at age 5 did predict greater *social competence* with unfamiliar peers (d=0.15) at age 7. The direct effect of *behavioral inhibition* on *social competence* was not significant, but the relationship between *behavioral inhibition* and *emotion regulation* (d=0.17) indicated that *emotion regulation* is the mechanism by which *behavioral inhibition* influences later *social competence*, albeit only among behaviorally inhibited children (d=0.19).

The relationship between temperament and social outcomes, as measured by the Infant Toddler Social-Emotional Assessment (ITSEA; a parent-reported measure of social-emotional difficulties and competencies), was examined in typically developing infants by Van Hecke et al. (2007). *Social fearfulness, inhibitory control, low pleasure,* and *soothability* was measured using the TBAQ-R at 15 months of age and social communication outcomes were examined via the *social competence* domain of the ITSEA at 2.5 years of age. Similar to Thompson and Lamb (1982) and Penela et al. (2015), early temperament was directly related to *social competence* at 2.5 years, being positively predicted by 15-month *inhibitory control* (d=0.87) and *low pleasure* (d=0.52).

Salley et al. (2013) examined the relationship between infant temperament and social responsiveness using the Social Responsiveness Scale (SRS). *Surgency/extraversion, negative affect,* and *effortful control* constructs were measured using the ECBQ at 2 years of age and social responsiveness was rated on the SRS subdomains of *social awareness, social cognition, social communication*, and *social motivation* at age 4. *Negative affect* at age 2 was the only temperamental dimension that was associated with overall social responsiveness, driven by *social cognition* (d=-1.32), *social communication* (d=-3.53), and *social motivation* (d=-1.86).

Salley et al. (2013) also compared the temperamental constructs of *surgency/extraversion, negative affect,* and *effortful control* at age 3 to later social responsiveness. Temperament was measured using the CBQ at 3 years and compared to ratings of social responsiveness on the SRS subdomains of *social awareness, social cognition, social* 

*communication*, and *social motivation* at 4 years. Similar to above, *negative affect* at age 3 was linked to greater impairments in *social communication* (d=-1.19) and *social cognition* (d=-2.08). Further, associations between *surgency/extraversion* and social responsiveness emerged at 3 years, where *surgency/extraversion* was associated with impairments in *social motivation* (d=-3.37) and *social communication* (d=-1.16). Positive associations between *effortful control* and *social awareness* were not evident until age 4 (d=-1.67).

#### Studies using the McDevitt Temperament Questionnaires

McDevitt Questionnaires were used in three studies to assess temperament during the infant and toddler periods (Houck, 1999; Bohlin et al., 2005; Hwang et al., 2009).

Houck (1999) investigated the relationship between infant temperament and the Adaptive Social Behavior Inventior (ASBI), specifically, the longitudinal relationship between *temperamental difficulty*, developmental competence, self-concept, and *social competence*. Infant *temperamental difficulty* was measured at 8 months using the ITQ-Revised (i.e., activity, adaptability, approachability, mood, and intensity subscales) and again at 12 months and 2 years of age using the TTQ. To measure *social competence*, the ASBI (express, comply, disrupt subscales) was used at 12 months, and again at 2 and 3 years. Overall, Houck (1999) found that *temperamental difficulty* at each time point was negatively related to *social competence*. Moreover, *temperamental difficulty* at 8 and 12 months, and 2 years of age was significantly correlated with *social competence* scores at subsequent time points. Effect sizes ranged from moderate to large (*d's=*-1.07 to -0.47).

Bohlin et al. (2005) investigated the relationships between infant temperament and Social Competence Inventory and Social Behavior Checklist to examine the interplay of infant *behavioral inhibition* and attachment security on the development of school-age *social competence*. *Behavioral inhibition* was derived from direct observations during infant approach by a friendly female stranger and parental ratings on the *approach-withdrawal* scale from the TTQ between 13 and 15 months of age, and attachment security was measured at 15 months. *Social competence with peers* was assessed at 8 years of age using an aggregate score of observations and ratings on the Social Competence Inventory and Social Behavior Checklist. *Behavioral inhibition* at 13-15 months did not predict *social competence* at 8 years (d=0.04). Interestingly, attachment security moderated the relationship, with a small effect size (d=0.28).

That is, *behavioral inhibition* in insecure infants negatively predicted later *social competence*, whereas it positively predicted later *social competence* for secure infants.

Hwang and colleagues (2009) examined the effects of early *temperament* on the development of *social-adaptive* outcomes at toddler and preschool ages using the Comprehensive Developmental Inventory for Infants and Toddlers (CDIIT). Toddlers were assessed between 18 months and 3 years to determine their early *temperament type* (i.e., easy, intermediate, or difficult) using the Chinese version of the TTQ. Their *social-adaptive* development was measured between 4 and 6 years using the *social-adaptive* factor of the CDIIT. Similar to Bohlin et al. (2005), toddler *temperament type* did not have a relationship with preschool *social-adaptive* outcomes (d=0.32).

### Studies using the Emotionality Activity Sociability Temperament Questionnaire

Two studies (Abulizi et al., 2017; Cha, 2017) used the Emotionality Activity Sociability (EAS; Buss & Plomin, 1984).

Abulizi et al. (2017) examined the relationship between the EAS and the Strengths and Difficulties Questionnaire (SDQ) to explore the longitudinal contribution of early temperamental traits on later behavioral problems in a sample of mother-child dyads. The EAS was used to assess *emotionality, activity, shyness,* and *sociability* at 12 months of age and the SDQ was used to measure a range of behavioral problems, including *peer-relation problems* at 5.5 years of age. Both *active* temperament (d=0.14) and *sociability* (d=0.14) in infancy were negatively correlated with *peer-relation problems* at 5.5 years. Moreover, Abulizi et al. (2017) further explored the associations between the temperamental traits and children's behavior; when adjusted for child's sex, premature birth, birth weight and birth order, family income, social support, and a range of parental covariates, infant *active* temperament predicted fewer *peer-relation problems* in childhood, with a moderate effect size (d=0.59).

The longitudinal interplay between *negative emotionality*, parenting, and early sociocognitive development was explored by Cha (2017), who explored the relationship between the EAS and the Korean Ages and Stages Questionnaire (ASQ) in a sample of Korean children. *Negative emotionality* was assessed using the emotionality scale of the EAS and socio-cognitive development was measured using the *communication* scale (expressive and receptive language development, communication orientation) and the *personal-social* scale (self-help skills and interactions with people or objects) of the ASQ. Both measures were administered at 4-5 months, 13-15 months, and 25-27 months. Overall, Cha (2017) reported no significant association between *negative emotionality* at 13-15 months or 25-27 months on subsequent *communication* and *personal-social* outcomes. However, *negative emotionality* at 4-5 months only marginally predicted low levels of *personal-social* outcomes at 13-15 months (d=0.10).

#### **B. Studies using Observational Measures of ER**

Seven studies used observational measures of ER and examined their relationship with later social communication, as described in Tables 3 and 4.

### Studies using the NICU Network Neurobehavioral Scale

The earliest time point in which ER was assessed observationally was at four-five weeks following birth, using the NICU Network Neurobehavioral Scale (NNNS; Bowers et al., 2018). Bowers et al. (2018) wanted to determine if infant measures of neurobehavior, specifically infant *arousal, self-regulation, excitability,* and *stress,* could predict later social and communicative behavior. At four to five weeks, the NNNS was administered and infants were followed longitudinally and assessed for *social behavior* using the SRS (a measure of social ability and impairment that is primarily used to measure autism severity) at 4, 5, and 8 years. Although the authors reported no significant associations between early *arousal, self-regulation, excitability, or stress* and *social behavior* at any of the SRS comparisons timepoints, there was a moderating effect with advanced maternal age, resulting in positive associations between *self-regulation* (*d*=0.37) and *arousal* (*d*=0.34) with *social behavior*, and negative associations between *self-regulation* (*d*=0.36) and *social behavior* at 5 years of age.

#### Studies using the Gentle Arm Restraint Task

Using the Gentle Arm Restraint task, Henderson (2007) examined the influence of early *negative emotional reactivity* on *social competence*. *Negative emotional reactivity* (calculated as the time spent in moderate/high distress during the task) was measured at 2 years and *social competence* was assessed using teachers' reports on the Preschool Socio-affective Profile (PSP) at 3 years. Overall, Henderson (2007) reported no direct correlation between *negative emotional reactivity* and *social competence* (d=0.47), but this relationship was significant when mother's

supportive parenting interacted with children's *negative emotional reactivity* (d=1.25). That is, when mothers exhibited high levels of supportive parenting, *negative emotional reactivity* was negatively correlated with later *social competence*, thereby promoting prosocial behavior (d=0.98).

#### Studies using the Two Bags Task

Rispoli et al. (2013) conducted a large-scale study of child social development – from infancy to kindergarten entry – examining the interrelations between parental behavior, attachment security, and child temperamental negativity, as well as their associations with later social competence. *Child negativity* was measured using the Two-Bags Task at 2 years and preschool age (~4 years) and social competence was measured twice during kindergarten (at ~5.5 and ~6 years), using items derived from the Preschool and Kindergarten Behavior Scales (PKBS-2), Social Skills Rating System (SSRS), Early Childhood Longitudinal Study – Kindergarten Class Social Rating Scale (ECLS-K), and Family and Child Experiences (FACES) social skills and positive approaches to learning scales. A significant relationship between *child negativity* and *social competence* at kindergarten entry (d=0.20). Conversely, in line with the Henderson (2007) study, 2-year *child negativity* was not related to *social competence* at kindergarten entry (d=0.12).

#### Studies using the Ainsworth Strange Situation Procedure

Belskey et al. (2001) investigated if infant *negative emotionality* could predict subsequent *social competence*, and if attentional persistence moderates this longitudinal relationship. *Negative emotionality* during the Strange Situation procedure and level of attention (during toy play) were measured at 15 months of age, followed by *social competence* using the Adaptive Social Behavior Inventory (ABSI) at 3 years. Infant *negative emotionality* was not significantly related to 3-year *social competence* (d=0.06). However, there was an effect of *negative emotionality* on later *social competence* as a function of infant attention level, where high levels *negative emotionality* predicted lower levels of *social competence* at 3 years, but only in children classified as having poor attentional persistence. Although a non-significant effect size (d=0.06),

high levels attention modestly regulated the effect of early *negative emotionality* on later *social competence*.

## Studies using Observational Measures of Affect Dysregulation

The NICHD Early Child Care Research Network (2004) conceptualized the construct of *affect dysregulation* in the toddler years by examining child and maternal antecedents contributing to early *affect dysregulation*, as well as its socio-emotional and cognitive consequences. At 6 months, infant *negative mood* and *difficult temperament* was assessed during the caregiver-child interaction measures in the home and the ITQ, respectively. At 2 and 3 years, *affect dysregulation* was measured using child-caregiver interactions (i.e., toy clean-up and play interaction) and children were assigned to an affect dysregulation group based on their level of negative affect or defiance. Maternal- and teacher-reported *social skills* were rated using the Social Skills Rating System (SSRS; i.e., cooperation, assertion, self-control, responsibility) at 4.5 years, and during kindergarten and grade 1. Overall, children with *affect dysregulation* at 2 and 3 years were less cognitively and socially competent at 4.5 years, kindergarten, and grade 1. That is, *affect dysregulation* at 2 years (but not 3 years) was associated with maternal rating of *social skills* across all time points, where mothers reported fewer *social skills* (*d*=0.30). Teachers also indicated that children in the *affect dysregulation* group at both 2 and 3 years showed fewer *social skills* from 4.5 years to grade 1 (*d*'s=0.22-0.24).

#### Studies using the Face-to-Face Still-Face Paradigm (FFSFP)

Two studies (Cassel, 2008; Parlade et al., 2009) utilized the Face-to-Face Still-Face Paradigm (FFSFP; Tronick et al., 1978).

Cassel (2008) investigated if early emotional expressions during the FFSFP were associated with subsequent joint attention and related competencies. Infant *smiles, cry-faces, neutral affect, socially directed expressions,* and *affective valence* were coded at 6 months and indices of joint attention – including *initiating joint attention (IJA*; as well as *anticipatory, reactive,* or *no smiling* during IJA episodes), *responding to joint attention (RJA),* and *initiating behavioral requests (IBR)* – were examined at 8, 10, 12, 15, and 18 months using the Early Social Communication Scales (ESCS). Emotional expressions during the FFSFP did not predict *IJA* or *RJA,* but were associated with *smiling* (during IJA episodes) and related competencies.

*Cry-faces* at 6 months were positively associated with *IBR* (d=1.28) and *reactive smiling* (d=3.37) at 8 months of age. Moreover, 6-month *neutral affect* was negatively correlated with *reactive smiling* at 8 (d=2.41) and 12 months of age (d=1.96). Similarly, *neutral affect* was also correlated with *no smiling* at 18 months (d=1.81), where infants exhibiting fewer expressions at 6 months showed even fewer during the ESCS at 18 months of age.

These findings were corroborated by Parlade and colleagues (2009), who examined the predictive relationship between *still-face smiling* and socially expressive behaviors during the ESCS. Proportion of time spent smiling during the still-face/unresponsive episode of FFSFP (i.e., *still-face smiling*) at 6 months of age was measured and compared to *IJA*, and *reactive* and *anticipatory smiling* (during IJA episodes) at 8, 10, and 12 months. In line with Cassel (2008), *still-face smiling* at 6 months was not significantly related to the rate of *IJA* at later ages. However, *still-face smiling* was positively associated with *anticipatory smiling* during joint attention episodes at 8 (d=1.42) and 10 months (d=1.15). That is, 6-month infants who used smiling/positive emotional expression to re-engage their caregiver during the still-face episode of the FFSFP were more likely to use anticipatory smiling to engage an unfamiliar social partner in object play during the ESCS at later ages. Significant and non-significant findings ranged from mid-level to large effect sizes.

# 2. Relationship Between Early Social Communication-Related Constructs and Subsequent ER

## A. Studies Using Observational Measures of Social Communication-Related Constructs

Just as ER influences the emergence of later social communication, studies have also examined antecedent social behaviors that may influence subsequent emotional development. When examining the directional relationship between social communication and subsequent ER, no studies in this review used questionnaire measures of social communication. Rather, four studies used observational measures to assess social communication. As shown in Table 2.3 and 2.4, these observational social communication measures included: the Face-to-Face Still-Face Paradigm, the Early Social Communication Scales and, the Stranger Sociability Procedure.

#### Studies using the Face-to-Face Still-Face Paradigm

Bedford et al. (2017) investigated if infant's mother-directed *gaze* could predict later *callous unemotional behaviors*. At 6 months of age, the FFSFP was administered to measure infant's mother-directed *gaze*. The infants were followed longitudinally and were assessed for emotion-related behavior, including facial *emotion recognition* using the Assessment of Children's Emotional Skills at 6 years and *callous unemotional behaviors* using the Inventory of Callous Unemotional Traits at 7 years. The authors reported no significant associations between infant *gaze* and *emotion recognition* (d=0.04) or *callous unemotional behaviors* (d=0.04), nor were there any indirect effects of infant *gaze* on *callous unemotional behaviors* via *emotion recognition* (d=0.47). There was, however, a moderating effect with maternal sensitivity, resulting in a negative relationship between infant *gaze* and later *callous unemotional* behaviors (d=0.50), but only for infants with low maternal sensitivity (i.e., infants with low maternal sensitivity and low levels of *gaze* showed higher levels of *callous unemotional behaviors* at age 7).

## Studies using the Early Social Communication Scales

Two studies (Van Hecke et al., 2007; Parlade et al., 2009) utilized the Early Social-Communication Scales (ESCS; Mundy et al., 2003) to measure nonverbal communication skills and social orienting behavior.

Van Hecke and colleagues (2007) examined the relationship between joint attention and later temperament; *IJA* (including high-level behavior), *RJA*, and *IBR* (including high-level behavior) were measured using the ESCS at 12 months of age, and temperamental outcomes using the TBAQ-R (*social fearfulness, inhibitory control, low pleasure,* and *soothability*) were examined at 15 months. Joint attention at 12 months was related to temperament at 15 months, with *RJA* negatively predicting *inhibitory control* (d=0.70), and high-level *IBR* negatively predicting *soothability* (d=0.75). Just as early temperament predicted social outcomes, it appeared that early joint attention predicted later temperament – potentially suggesting a bidirectional association between the developmental systems.

A follow-up study by Parlade et al. (2009) found that IJA measures predicted socioemotional outcome as indexed by the ITSEA. Using a subsample of participants from the Van Hecke (2007) study, *IJA, anticipatory,* and *reactive smiling* was measured during the ESCS at 9 and 12 months. Social-emotional outcome, using the ITSEA *dysregulation, externalizing,* and *internalizing* domains, was assessed at 2.5 years of age. Interestingly, ITSEA *dysregulation, externalizing*, and *internalizing* domains were not significantly related to any *IJA* or *smiling variables* at 9 (d's=0.02 to 0.58) or 12 months (d's=0.08 to 0.54).

#### Studies using the Stranger Sociability Procedure

Thompson and Lamb (1982) assessed the associations between *sociability* and temperament. Using the Stranger Sociability procedure, infant *sociability* was measured at 12.5 months of age, followed by measurement of temperament (i.e., *activity level, distress to limitations, fear, duration of orienting, smiling and laughter,* and *soothability*) at 19.5 months using the IBQ. Interestingly, *sociability* was negatively associated with *fear* (d=0.82) – consistent with the inverse relationship. Taken together, it appeared that stranger *sociability* is implicated in the development of *fearful* temperament, irrespective of direction.

#### DISCUSSION

The present review provided an in-depth overview of research that has examined the bidirectional developmental relationships between ER and social communication in childhood. The review had three main findings. First, a narrative synthesis revealed an age-related pattern, where predictive bi-directional relationships emerged following six months of age between ER and social communication. Second, there was tremendous heterogeneity among studies when defining ER constructs and methodology used to evaluate ER. Third, similar to ER, there was heterogeneity among social communication studies, with respect to constructs and assessment measures. While this review illustrates that children's regulation of emotions is essential to the development of social communication and vice versa, there is little consensus in the field concerning direct relationships due to heterogeneity, as well as a lack of consistency and replication across studies.

There were no direct relationships from ER to social communication, or social communication to ER when first assessed between the ages of one and six months using questionnaires and behavioral assessments. Predictive relationships generally appeared after six months of age, with different questionnaires and observational measurements of ER predicting social communication constructs and vice versa. There are a few exceptions to this pattern of findings. For example, studies that used an author-created ER construct (combining two or more

questionnaire subscales) did not find a significant relationship, which may have resulted from washing out the effects of the individual subscales when combined (Henderson et al., 2001; Penela et al., 2015). Similarly, instances of initiating joint attention and smiling did not predict internalizing and externalizing composites (Parlade et al., 2009), supporting a washed out effect for composite scores. Further, the significant findings in this review stem from relationships between social communication constructs and ER subscales, rather than ER composites. Finally, significant relationships were found in six studies only when moderators were included in the analyses (Bedford et al., 2017; Belskey et al., 2001; Bohlin et al., 2005; Bowers et al., 2018; Henderson, 2007; Penela et al., 2012), suggesting that the relationship between ER and social communication may be more complex and nuanced than a direct one-to-one relationship.

To date, historical and empirical literature have addressed the controversial topic of how to best define and measure ER, and the current review further illuminates these complexities. There was tremendous heterogeneity across studies when defining and measuring ER. There were a total of 17 questionnaire or observational measures used to index ER, where many studies employed more than one measure. Of these, 30 ER constructs were assessed: arousal, activity level, regulation, dysregulation, excitability, stress, laughter, smiling, fear, distress to limitations, soothability, duration of orienting, cuddliness, perceptual sensitivity, low-intensity pleasure, inhibition, negative emotionality, positive emotionality, surgency, extraversion, effortful control, negative affect, positive affect, temperamental type/difficulty, shyness, sociability, reactivity, emotional skills/traits, internalizing behavior, and externalizing behavior. There was general consensus across studies that ER is an individual-context transactional process and involves strategies to control, modify, and manage aspects of a child's response to emotioneliciting stimuli. Moreoever, studies shared the perspective that ER contributes to the formation and maintenance of social communication - including engaging in prosocial relationships, effective social communication, social competence, decision-making, and attention (Calkins, 1994; Calkins & Hill, 2007; Cole et al., 2004; Eisenberg et al., 1995; Kopp, 1982; Thompson, 1994). However, interpretation of findings across studies was difficult for three reasons. First, the operationalization of ER, including using singular or multiple constructs and/or the examination of theoretical frameworks or models to assess ER (such as temperament theory, Connor-Smith et al. (2001) ER framework, or Gross & Thompson's (2007) modal model of ER) resulted in different organizing matrices, further increasing the complexity of findings and

difficulties in comparing across studies. Second, different methodologies used (e.g., observational or informant report, single or multi-method assessment, etc.) was also considered. A review of 35 years of ER research in typical-developing individuals reported that while majority of published research examined more than one ER construct, 61.1% of studies employed a singular versus a multi-methodological approach (Adrian et al., 2011), consistent with findings in the present review. Finally, factors including maturational processes, social environment, and temperament (which comprised of 15 studies in the current review) influence ER capacities, further tapping into the nuances of the ER process. Overall, ER is a complicated, multifaceted process and there appears to be little consensus in the field in how to best conceptualize and measure ER in typical-developing children, including the relation to social communication outcomes.

In parallel with ER, there was heterogeneity among studies when defining and measuring social communication, rendering comparison and replication of findings difficult. There were a total of 21 questionnaires or observational measurements of social communication. Of these, 17 social communication constructs were assessed: social skills, social behavior, social interaction, social engagement, social competence, sociability, social adaptation, social cognition, social motivation, social awareness, responsiveness, involvement, aggression, gaze, joint attention, *peer relationships*, and *smiling*. A limitation in the present review was utilizing a term that encompassed all constructs related to social communication. Given that many aspects of a child's social functioning fall under the mantle of social communication, we chose constructs that mapped onto skills and behaviors used in reciprocal social interactions. This broadly included skills/behaviors related to verbal and non-verbal language use in social exchanges, social awareness, social information processing and cognition, social motivation, and capacity for social interaction (Constantino, 2002). Because different types of social communication and numerous variants of those constructs (e.g., social skills, social behavior, sociability) were included, organizing and interpreting data across studies was complex. Moreover, similar to the operationalization of ER, intrinsic (e.g., biological processes and temperament) and extrinsic (e.g., social network, parental attachment, etc.) factors may have further impacted the generalizability of social communication findings.

Assessing and understanding the developmental relationships between ER and social communication are important, as these skills form the foundation for later functioning –

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including school success, building lasting relationships with family and friends, and navigating the social world (Pontoppidan, 2017). When developmental milestones for ER and social communication are met, a child is able to experience, manage, and express a full range of emotions; become aware of others' emotions and social cues; develop satisfying relationships with other peers and adults; cope effectively with aversive or challenging interactions; develop emotional communication and empathy within relationships; and actively explore and engage with their environment and learn (Saarni, 1999). Thus, it seems likely that a child's ability to regulate their emotions and navigate social exchanges interact in a synergistic fashion, allowing for harmonious interpersonal interactions. On the other hand, when milestones for ER and social communication are not met, or if the developmental trajectory goes amiss, deficits in these constructs are likely to emerge. This may contribute to atypical social-emotional outcomes, including increased risk for mental health disorders or neurodevelopmental disorders (McElwain, 1999). As such, examining ER and social communication in tandem to determine how they influence children's behavior and relationships is imperative. Interestingly, to date, the vast majority of research examines these constructs independently, are hypothesis-driven, focus on moderating or mediating factors (i.e., indirect effects), and/or investigates one directional relationship and not the other. In fact, our narrative synthesis revealed that the majority of studies examined the influence of early ER on later social communication (n=19), while a paucity of studies examined the reciprocal relationship (social communication to ER; n=1) or bi-directional relationships (n=3). This may be attributed to the paucity of studies exploring how social communication predicts and influences later ER.

As with other literature reviews, the present review had a number of limitations. Although we strived to be inclusive during our search and included four databases without language or publication date restrictions, it is possible that articles may have been missed that were not indexed in our selected databases. As well, our search strategy included a combination of terms that combined concepts of emotion, emotion regulation, social communication, infancy, and longitudinal study design. It is possible that other studies were not ascertained by these keywords and subject headings. To address this limitation, future studies may consider additional search terms and include book chapters, literature reviews, or other publications that do not contain empirical data. As previously noted, there was a lack of consistency and replication across studies due to heterogenity in the operationalization of ER and social communication.

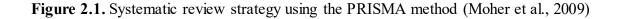
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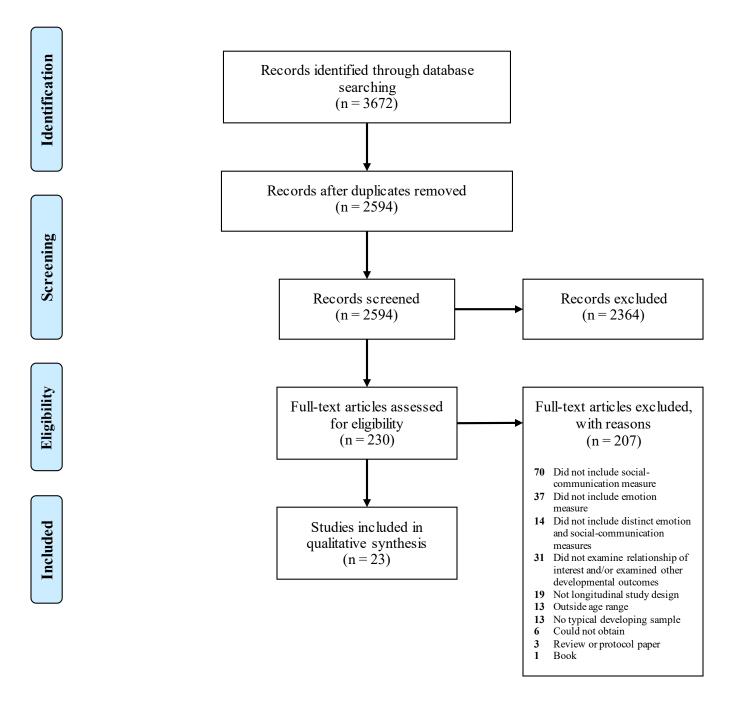
There was no one definition or assessment measure used consistently, rendering comparison across studies difficult and an inability to draw thorough conclusions about the bidirectional relationship between ER and social communication. Because of the heterogeneity among measures, it was not possible to conduct a meta-analyses to generate pooled effect estimates. Moreover, given that studies were of different sizes and the complexity of measurement models, power varied considerably such that statistical significance had variable interpretation across studies (e.g., some studies with small to moderate d's were not statistically significant, whereas others with much smaller d's were significant). Perhaps approaching the study of ER, social communication, and the relationship between the two from a specific theoretical framework would result in a different organizing matrix. That is, a theoretical framework of how ER maps onto social communication, and vice versa, and onto actual measures to assess these constructs may provide clarity. Careful theoretical unpacking and devising clearly differentiated measures of ER and social communication presents a challenge for future studies. Finally, as we wanted to fully capture the longitudinal relationship between ER and social communication (and vice versa), there was variability in the age ranges examined. While our inclusion criteria required at least one time point in infancy ( $\leq 2$  years of age), there was no restriction on the upper age limit. As a result, study ages ranged from as early as one month to eight years of age. This large scope/variability proved difficult when comparing studies longitudinally. Moreover, certain assessment measures were restricted for use at certain ages (e.g., IBQ for three to 12 months, TBAQ for 18 months to 3 years), again, rendering comparison between studies complex. Future studies may consider opting for a defined age range (e.g., infancy to toddlerhood), or selecting specific assessment measures to ensure methodological overlap.

#### **CONCLUSION**

This review summarized the results of research that has examined the developmental relationships between ER and social communication in childhood. Although there was a general age-related pattern that appeared following six months of age, where ER was predictive of subsequent social communication and vice versa, there was no consensus regarding the direct nature of these relationships due to heterogeneity and a lack of consistency across studies. While this review has documented the importance of ER and social communication and their contributions to developmental outcomes, devising clearly differentiated definitions and

measures of these constructs presents a challenge for future research. More attention is required to disentangle and deconstruct the conceptual and methodological issues in ER and social communication research. Understanding the potential causal relationships and pathways between ER and social communication is important and may inform future studies, both in typically developing children and children with atypical presentations.





# Table 2.1. Study Demographics

First Author and Year	Sample Size and Sex	Ages Assessed	Study Objective	Comparison Group, if applicable
		Studies Examining E	R → Social Communication Relationship (n=19)	•
Albers et al., 2007	<i>n</i> = 64 34M:30F	3m, 6m	Investigated if 3-month infant negative emotionality was associated with the quality of social interaction with professional caregivers in childcare centers at 6 month of age	N/A
Aureli et al., 2017	n = 80 41M:39F	4m, 6m	Investigated the relation between 4-month infant general temperament with mother-infant interactions during dyadic and triadic play at 6 months of age	N/A
Penela et al., 2012	<i>n</i> = 155 74M:81F	9m, 2 years	Investigated the influence of infant temperamental fear at 9 months on children's social interactions and engagement at 2 years of age	N/A
Henderson et al., 2001	<i>n</i> = 97 46M:51F	9m, 4 years	Investigated the influence of 9-month infant negative reactivity on measures of sociability at 4 years of age	N/A
Markova, 2008	<i>n</i> = 60 27M:33F	6m, 7m, 9m, 12m	Investigated the influence of 6-month infant general temperament on social competence with peers, specifically interaction skills and dyadic preferences, at 7, 9, and 12 months of age	N/A
Karass, 2002	<i>n</i> = 83 48M:35F	4m, 8m, 12m, 16m	Investigated the relations between 4, 8, 12, 16-month positive and negative emotionality with subsequent joint attention during mother-infant free play at 12 and 16 months of age	N/A
Penela et al., 2015	n = 257 122M:135F	2, 3, 5, 7 years	Investigated if 2- and 3-year temperamental behavioral inhibition was associated with 5-year ER strategies during a disappointing context task and 7-year social competence during free play with an unfamiliar peer	N/A
Salley et al., 2013	n = 60 31M:29F	2, 3, 4 years	Investigated the relations between temperament dimensions at 2 and 3 years with social responsiveness outcomes at 4 years of age	N/A
Houck, 1999	n = 126 85M:41F	8m, 12m, 2, 3 years	Investigated the relations between infant temperamental difficulty at 8 and 12 months, and 2 years with social competence at 12 months, and 2 and 3 years of age	N/A
Bohlin et al., 2005	<i>n</i> = 90 44M:46F	13-15m, 8 years	Investigated if 13 to 15 month temperamental behavioral inhibition and attachment security were associated with school-age social competence	N/A
Hwang et al., 2009	<i>n</i> = 110 N/A	18m-3 years, 4-6 years	Investigated the influence of temperament type during the toddler period (18 months to 3 years) on social-adaptive development during preschool age (4 to 6 years)	Pre-term infants
Abulizi et al., 2017	<i>n</i> = 1184 626M:558F	12m, 5.5 years	Investigated the contribution of 12-month temperamental traits on subsequent peer-relation problems at 5.5 years of age	N/A
Cha et al., 2017	<i>n</i> = 1620 828M:792F	4-5m, 13-15m, 25- 27m	Investigated the relations between infant negative emotionality and socio- cognitive development (i.e., communication and personal-social skills)	N/A

			during the first 2 years of life	
Bowers et al., 2018	<i>n</i> = 227 98M:129F	4-5 weeks, 4, 5, 8 years	Investigated if infant neurobehavior at 4-5 weeks could predict social and communicative behavior at 4, 5, and 8 years	N/A
Henderson, 2007	<i>n</i> = 55 20M:35F	2, 3 years	Investigated the influence of 2-year infant negative emotional reactivity during the Gentle Arm Restraint task and mothers' parenting style on social competence at 3 years of age	N/A
Rispoli et al., 2013	<i>n</i> = 6850 3473M:3377F	2, ~4 (preschool), ~5.5, ~6 (kindergarten) years	Investigated the associations between child negativity at 2- and 4 (preschool age) years during the Two-Bags Task with kindergarten social competence at 5.5 and 6 years of age	N/A
Belskey et al., 2001	<i>n</i> = 1088 N/A	15m, 3 years	Investigated if 15-month infant negative emotionality could predict social competence at 3 years of age, and if this relationship is moderated by attentional persistence	N/A
NICHD Early Child Care Research Network, 2004	<i>n</i> = 1023 518M:505F	6m, 2, 3, 4.5 years, kindergarten, grade 1	Investigated the contribution of 6-month temperament on toddler affect dysregulation at 2 and 3 years, and the subsequent effects on socio- emotional development and social skills at 4.5 years and during kindergarten and grade 1	N/A
Cassel, 2008	<i>n</i> = 17 8M:9F	6m, 8m, 10m, 12m, 15m, 18m	Investigated if 6-month infant emotional expressions and affective valence during the Face-to-Face/Still-Face Paradigm were associated with joint attention at 8, 10, 12, 15, and 18 months of age	High-risk infant siblings
		Studies Examining So	ocial Communication → ER Relationship (n=1)	
Bedford et al., 2017	n = 206 N/A	6m, 6, 7 years	Investigated if 6-month infant gaze during the Face-to-Face/Still-Face Paradigm could predict emotional recognition at 6 years and callous unemotional behaviors at 7 years of age	N/A
		Studies Exam	ining Bidirectional Relationships (n=3)	
Thompson et al., 1982	<i>n</i> = 43 21M:22F	12.5m, 19.5m	Investigated the bi-directional relationship between temperament and sociability during the Stranger Sociability procedure at 12.5 and 19.5 months of age	N/A
Van Hecke et al., 2007	n = 52 18M:34F	12m, 15m, 2.5 years	Investigated the bi-directional relationships between 12-month infant joint attention, 15-month general temperament, and later social competence at 2.5 years of age	
Parlade et al., 2009	Study 1: <i>n</i> = 26 13M:13F Study 2: <i>n</i> = 39 27M:33F	6m, 8m, 9m, 10m, 12m, 2.5 years	Investigated the bi-directional relationships between 6-month smiling/positive emotional expression during the Face-to-Face/Still-Face Paradigm, joint attention at 8, 9, 10, and 12 months, and social-emotional outcomes at 2.5 years	N/A

Article	ER Measure and Age Assessed	ER Construct/Domain(s)	Social Communication Measure and Age Assessed	Social Communication Construct/Domain(s)				
		Studies Examining ER → Social Co	ommunication Relationship					
ER Questionnair	R Questionnaires (n=15)							
Albers et al., 2007	<b>3m:</b> IBQ-Revised	Negative emotionality	<b>6m:</b> Professional caregiver-infant interactions (i.e., changing diapers, feeding, putting to bed)	Infant responsiveness Involvement				
Aureli et al., 2017	4m: IBQ-Revised	Cuddliness Soothability Perceptual sensitivity Low intensity pleasure Duration of orienting	<b>6m:</b> Mother-child interaction during face-to-face (dyadic) and object (triadic) play	Co-regulation (i.e., unilateral, asymmetrical, symmetrical, disruptive, unengaged)				
Penela et al., 2012	<b>9m:</b> IBQ	Fear	<b>2 years:</b> Free play with unfamiliar, same- sex peer	Social engagement Aggression				
Henderson et al., 2001	<b>9m:</b> IBQ	Negative reactivity	<b>4 years:</b> Free play with unfamiliar peers, CCTI	Sociability				
Markova, 2008	6m: IBQ	Negative affectivity Extraversion Regulation	7m, 9m, 12m: Peer group interaction with same-sex peers	Interaction skills (i.e., gaze, facial expressions, vocalizations, initiations, responses to initiations, coordinated attention, communicative gestures, touch, reach, play) Dyadic preferences for peers				
Karass, 2002	4m, 8m, 12m, 16m: IBQ	Positive emotionality (i.e., duration of orienting, smiling and laughter, soothability) Negative emotionality (i.e., activity level, distress to limitations, distress to novelty)	12m, 16m: Mother-infant free play	Joint attention				
Thompson et al., 1982	<b>12.5m:</b> IBQ	Activity level Distress to limitations Fear Duration of orienting	<b>19.5m:</b> Stranger Sociability Procedure	Sociability				

		Smiling and laughter		
		Soothability		
Penela et al., 2015	<ul><li>2, 3 years: TBAQ</li><li>5 years: Disappointment context paradigm</li></ul>	Behavioral inhibition Emotion regulation	7 years: Free play with unfamiliar peers	Social competence
Van Hecke et al., 2007	15m: TBAQ-Revised	Social fearfulness Inhibitory control Low pleasure Soothability	<b>2.5 years:</b> ITSEA	Social competence
Salley et al., 2013	2 years: ECBQ 3 years: CBQ	Surgency/extraversion Negative affect Effortful control	<b>2, 3, 4 years:</b> SRS	Social awareness Social cognition Social-communication Social motivation
Houck, 1999	8m: ITQ-Revised 12m, 2 years: TTQ	Temperamental difficulty	<b>12m, 2, 3 years:</b> ABSI	Social competence
Bohlin et al., 2005	<b>13-15m:</b> TTQ, Infant approach by friendly stranger	Behavioral inhibition	<b>8 years:</b> Social Competence Inventory, Social Behavior Checklist	Social competence
Hwang et al., 2009	18m-3 years: TTQ	Temperament type (i.e., early, intermediate, difficult)	4-6 years: CDIIT	Social-adaptive development
Abulizi et al., 2017	12m: EAS	Emotionality Activity Shyness Sociability	5.5 years: SDQ	Peer-relation problems
Cha et al., 2017	<b>4-5m, 13-15m, 25-27m:</b> EAS	Negative emotionality	<b>4-5m, 13-15m, 25-27m:</b> Korean ASQ	Communication development Personal-social development
<b>Observational Me</b>	asures of ER (n=7)			
Bowers et al., 2018	4-5 weeks: NNNS	Arousal Self-regulation Excitability Stress	4, 5, 8 years: SRS	Social behavior
Henderson, 2007	<b>2 years:</b> Gentle Arm Restraint task	Negative emotional reactivity	<b>3</b> years: PSP	Social competence
Rispoli et al., 2013	<b>2, ~4 (preschool) years:</b> Two Bags Task	Negativity	<b>5.5 and 6 (kindergarten) years:</b> PKBS-2, SSRS, ECLS-K, FACES, Positive	Social competence

			Approaches to Learning scale	
Belskey et al., 2001	<b>15m:</b> Strange Situation procedure	Negative emotionality	3 years: ABSI	Social competence
NICHD Early Child Care Research Network, 2004	<ul> <li>6m: ITQ, caregiver-child play interaction</li> <li>2, 3 years: caregiver-child play interactions</li> </ul>	Negative mood Difficult temperament Affect dysregulation	<b>4.5 years, kindergarten, grade 1:</b> SSRS (parent and teacher report)	Social skills (i.e., cooperation, assertion, self-control, responsibility)
Cassel, 2008	6m: FFSFP	Smiles Cry-faces Neutral affect Socially-directed expressions Affective valence	8m, 10m, 12m, 15m, 18m: ESCS	Joint attention (i.e., initiating joint attention, responding to joint attention, initiating behavioral requests)
Parlade et al., 2009	6m: FFSFP	Still-face smiling	8m, 10m, 12m: ESCS	Initiating joint attention Reactive smiling Anticipatory smiling
		Studies Examining Social Commu	nication → ER Relationship	
<b>Observational M</b>	easures of Social Communic	ation (n=4)		
Bedford et al., 2017	<ul> <li>6 years: Assessment of Children's Emotional Skills</li> <li>7 years: Inventory of Callous Unemotional Traits</li> </ul>	Emotion recognition Callous unemotional behaviors	6m: FFSFP	Gaze
Van Hecke et al., 2007	15m: TBAQ-Revised	Social fearfulness Inhibitory control Low pleasure Soothability	12m: ESCS	Initiating joint attention Responding to joint attention Initiating behavior requests
Parlade et al., 2009	2.5 years: ITSEA	Dysregulation Internalizing behaviors Externalizing behaviors	9m, 12m: ESCS	IJA Reactive smiling Anticipatory smiling
Thompson et al., 1982	<b>19.5m:</b> IBQ	Activity level Distress to limitations Fear Duration of orienting Smiling and laughter Soothability	<b>12.5m:</b> Stranger Sociability procedure	Sociability

Author	Construct	Measure Name	Description
Questionnaires			
Rothbart et al., 2001	ER	Children's Behavior Questionnaire (CBQ)	Designed to assess temperament in early to mid-childhood (aged 3 to 7 years). It is comprised of 15 subscales, including 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. Three factors evaluate broad dimensions of temperament: extraversion/surgency, negative affectivity, and effortful control. Items are scored on a 7-point scale ranging from 1 (never) to 7 (always).
Putnam et al., 2006	ER	Early Child Behavior Questionnaire (ECBQ)	Developed to provide a more comprehensive assessment of toddler temperament that indexes reactive processes involving emotion, motor and sensory systems, and self-regulatory processes. Measures temperament in children between 12 months and 3 years of age and is comprised of 18 scales, which are extensions of the dimensions contained on the IBQ and CBQ, and three scales from the TBAQ. Items are scored on a 7-point scale ranging from 1 (never) to 7 (always).
Buss & Plomin, 1984	ER	Emotionality Activity Sociability (EAS)	Parent-reported measure of childhood temperament. The EAS assesses four temperamental dimensions: emotionality (irritability/anger), activity (activity level), shyness (fear), and sociability (positive affect, including approach). It is recommended for children aged 12 months to 9 years and consists of 20 items, rated on a 5-point scale ranging from 1 (my child's behavior is never like this) to 5 (my child's behavior is always like this). The items belonging to each temperament are scored to form the four temperament dimensions.
Rothbart, 1981	ER	Infant Behavior Questionnaire (IBQ)	Assesses temperament in infants between 3 and 12 months of age. The IBQ is comprised of six subscales, including activity level, smiling and laughing, fear, distress to limitations, soothability, and duration of orienting. Items are scored on a 7-point scale ranging from 1 (never) to 7 (always), and an 8 <sup>th</sup> point that does not apply. The IBQ is well-validated and possessed excellent test-retest reliability (Goldsmith & Rothbart, 1991).
Gartstein & Rothbart, 2003	ER	Infant Behavior Questionnaire - Revised (IBQ-R)	Designed to assess temperament in children aged 3 to 12 months and has fourteen subscales: activity level, smiling and laughing, fear, distress to limitations, high pleasure, low pleasure, soothability, falling reactivity, cuddliness, sadness, approach, vocal reactivity, perceptual sensitivity, and duration of orienting. It can be completed by parents within 15 minutes, is well validated, and has excellent test-retest reliability (Goldsmith & Rothbart, 1991).
Carey & McDevitt, 1978	ER	Infant Temperament Questionnaire (ITQ)	Designed to assess temperament in infants from 4 to 8 months of age. Consisting of 95 items, the ITQ examines several areas of behavior, including responses to people and the environment, feeding, sleeping, dressing, diapering, and bathing. Items are scored on a 6-point scale from 1 (almost never) to 6 (almost always). A difficulty score can also be calculated from five subscales: adaptability, approachability, activity, mood, and intensity.

# Table 2.3. Descriptions of Questionnaires and Observational Measures

Goldsmith, 1996	ER	Toddler Behavior Assessment Questionnaire (TBAQ	Measures temperament in toddlers between 18 months and 3 years of age. It is comprised of five scales and measures the temperamental dimensions of activity level, anger proneness, interest/persistence, pleasure, and social fearfulness. Items are scored on a 7-point scale ranging from 1 (never) to 7 (always).
Rothbart et al., 2003	ER	Toddler Behavior Assessment Questionnaire - Revised (TBAQ-R)	A reliable and well-validated measure of temperament designed for infants 18 to 35 months. It is comprised of 13 scales, including the original scales from the TBAQ, to measure activity level, inhibitory control, anger, interest, pleasure, object and social fear, soothability, attention, sadness, and perceptual sensitivity. Items are scored on a 7-point scale ranging from 1 (never) to 7 (always).
Fullard et al., 1979	ER	Toddler Temperament Scale (TTQ)	Designed to assess temperament in toddlers aged 12 months to 3 years. Consisting of 97 items scored using a 6-point scale from 1 (almost never) to 6 (almost always). A difficulty score can also be calculated.
Observational Me	easures		
NICHD Study of Early Child Care, 2004	ER	Affect Dysregulation	Two play-based observations of caregiver-child interaction: toy clean-up activity and semi-structured play interaction. The lab clean-up tasks indexes defiant non-compliance and negative affect, and involves the caregiver and child participating in picking up toys. Child behaviors, including compliance, assertive noncompliance, defiant noncompliance, positive affect, and negative affect, are scored on a 5-point scale from 1 (not characteristic) and 5 (very characteristic). The caregiver-child play interaction measures child negative mood and negativity, and involves a three-box procedure where the caregiver is instructed to show their child age-appropriate toys in three containers in a set order. Maternal behaviors, including sensitivity to non-distress/supportive presence, intrusiveness/respect for child's autonomy, detachment, stimulation of cognitive development, positive regard for the child, negative regard for child/hostility, flatness of affect, and confidence are scored on a 4-point scale at 2 years, and 7-point scale at 3 years. Child behaviors, including positive mood/enthusiasm, negative mood/negative persistence, activity level, sustained attention, and engagement/affection with caregiver, are scored on a 4-point scale.
Mundy et al., 2003	Social Communication	Early Social- Communication Scales (ESCS)	An examiner and child are seated facing each other at a small table, where the child is presented with standardized presses (e.g., using active wind-up toys, hand-operated toys, ball, book with large distinct pictures, colorful posters on the walls to the left, right, and behind the child, etc.) that provide opportunities for social interaction, joint attention, and/or behavioral requests. The ESCS classifies children's behavior into three categories of early social communication behavior: a) joint attention (nonverbal behavior to share the experience of events or objects with others using eye contact, pointing, and showing; includes initiating and responding to joint attention), b) behavioral requests (nonverbal behavior to elicit help in obtaining events or objects; includes initiating and responding to behavioral requests), and c) social interaction behavior (child's ability to engage in playful, affective turn-taking interactions with others; includes initiating and responding to social interactions).

Tronick et al., 1978	ER and Social Communication	Face-to-Face Still-Face Paradigm (FFSFP)	Observational assessment of socio-emotional regulation comprised of three episodes: face-to-face, still- face, and reunion. Caregivers are situated at a table directly across and in front of their infant, and given a set of standardized instructions for each episode. During the face-to-face, caregivers are instructed to play with their infants as they normally would for 2 min (play episode). This is followed by the still- face episode, where caregivers are instructed to stop any facial or verbal communication directed to their infant and maintain a still face while keeping eye contact for 2 min (still-face episode). After the still-face episode, the caregiver resumes normal, face-to-face play with their infant for 2 min (reunion episode).
Goldsmith et al., 1999	ER	Gentle Arm Restraint Task	Caregivers sit behind their child on the floor. The child is presented with an attractive musical toy. After 30 seconds of playing, caregivers are instructed to hold their child's arms gently, but firmly to his/her sides (and ensure that the child cannot break free). Following a 30 second restraint, caregivers release their child's arms and allow play with toy to resume. This sequence is generally repeated twice.
Lester et al., 2004	ER	NICU Network Neurobehavioral Scale (NNNN)	Standardized assessment designed to measure multiple aspects of neurobehavior in typical and at-risk infants, including central nervous system integrity, neurological function, sensory/ interactive responses, and signs of stress
Ainsworth & Wittig, 1969	ER	Strange Situation procedure	Designed to evoke and measure negative emotion and attachment security in children aged 12 months to 2 years, the Strange Situation procedure comprises of a series of 8 episodes of approximately 3 min each: 1) caregiver, child, and experimenter in room, 2) caregiver and child alone, 3) stranger joins caregiver and child, 4) caregivers leaves and child is alone with stranger, 5) caregiver returns and stranger leaves, 6) caregiver leaves and child is left alone in room, 7) stranger returns and child is alone with stranger, and 8) caregiver returns and stranger leaves. The degree of negative emotion and distress exhibited by the child is rated on a 5-point scale by examining exploratory behaviors (e.g., moving around room), search behaviors (e.g., following caregiver to door), and affect displays (e.g., crying or smiling). As well, attachment classifications can also be derived based on interaction behaviors (proximity and contact seeking, maintaining contact, avoidance of contact and proximity, and resistance to contact and comforting) directed to the caregiver during the two reunion episodes.
Stevenson & Lamb, 1979	Social Communication	Stranger Sociability procedure	Designed to measure an infant's social-emotional responses and social initiatives to overtures with an unfamiliar adult. The procedure involves a series of social encounters or bids with increasing intrusiveness by an unfamiliar adult (in the presence of the caregiver). First, while the infant is seated on the caregiver's lap, the unfamiliar adult offers the infant an interesting toy, followed by attempts to initiate a give-and-take exchange. Subsequently, the infant is placed on the floor and the initial behavioral response is observed. The unfamiliar adult moves to the floor and offers another the infant another toy, followed by attempts to initiate turn-taking. The unfamiliar adult then attempts to pick up the infant, before leaving the room. The infant's initial behavioral response to each social overture is scored on a 5-point scale from 1 (withdrawal/distress) to 5 (friendly and outgoing).

Rispoli et al., 2013	ER	Two Bags Task	Adapted from the Three Bags Task used in the Early Head Start Research and Evaluation Project (Love et al., 2005) and in the NICHD Early Child Care Research Network (Owen et al., 1996). Designed to elicit caregiver and child socio-emotional behavior, this semi-structure task contains two bags with two different sets of developmentally appropriate toys (e.g., toys for pretend play and joint book reading). During the task, the caregiver and child are instructed to play for 10 min with items from the two bags, beginning with the first bag before switching to the second bag. Caregiver-child interactions are video-taped and coded for parental, child, and dyadic behaviors and emotions. There are three child-rating global scales (child engagement of caregiver, child negativity towards caregiver, and child quality of play/sustained attention) and six caregiver-rated scales (parental sensitivity, parental intrusiveness, cognitive stimulation, parental positive regard, parental negative regard, and parental detachment), each rated on a 7-point scale from 1 (very low) to 7 (very high).
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# Table 2.4. Descriptions of Results

Article	ER Construct/Domain(s) and Age Assessed	Social Communication Construct/Domain(s) and Age Assessed	Significant Differences	Effect Size (Cohen's <i>d)</i>	Significant Relationship(s) Between Constructs/Domains
		Studies Examini	ng ER → Social Communication	n Relationship	
ER Questionnaire	es (n=15)				
Albers et al., 2007	<b>3m</b> Negative emotionality	<b>6m</b> Infant responsiveness Involvement	No significance	No significance: d's=0.06	N/A
Aureli et al., 2017	<b>4m</b> Cuddliness Soothability Perceptual sensitivity Low intensity pleasure Duration of orienting	<b>6m</b> Co-regulation (i.e., unilateral, asymmetrical, symmetrical, disruptive, unengaged)	Soothability vs. symmetrical co- regulation	Moderate: <i>d</i> =0.36	↑ soothability = ↑ symmetrical co- regulation
Penela et al., 2012	<b>9m</b> Fear	<b>2 years</b> Social engagement Aggression	Fear vs. social engagement (moderated by low maternal care) Fear vs. aggression (moderated	Small: <i>d</i> =0.20	<ul> <li>↑ fear = ↓ social engagement (moderated by low maternal care)</li> <li>↓ fear = ↑ aggression (moderated by low</li> </ul>
			by low maternal care)	<i>d</i> =0.17	maternal care)
Henderson et al., 2001	<b>9m</b> Negative reactivity	<b>4 years</b> Sociability	No significance	Moderate: <i>d</i> =0.35	N/A
		7m, 9m, 12m Interaction skills (i.e., gaze, facial	i. Negative affectivity vs. coordinated attention	Small: <i>d</i> =0.47	↑ negative affectivity = $\downarrow$ coordinated attention
Markova, 2008	<b>6m</b> Negative affectivity	expressions, vocalizations, initiations, responses	ii. Extraversion vs. age vs. communicative gestures (12m)	Moderate: <i>d</i> =0.52	$\uparrow$ extraversion = $\uparrow$ communicative gestures (12m)
	Extraversion Regulation	to initiations, coordinated attention, communicative	iii. Negative affectivity vs. dyadic preferences (9m)	Small: <i>d</i> =0.42	↑ negative affectivity = ↓ dyadic preferences (9m)
			iv. Extraversion vs. dyadic preferences (12m)	No significance: <i>d</i> =0.06	$\uparrow$ extraversion = $\uparrow$ dyadic preferences (12m)

		Dyadic preferences for peers			
Karass, 2002	<b>4m, 8m, 12m, 16m</b> Positive emotionality (i.e., duration of orienting, smiling and laughter, soothability) Negative emotionality (i.e., activity level, distress to limitations, distress to novelty)	<b>12m, 16m</b> Joint attention	No significance	Small: <i>d's</i> =0 -0.32	N/A
Thompson et al.,	<b>12.5m</b> Activity level Distress to limitations	19.5m	Activity level vs. sociability Distress to limitations vs.	Moderate: <i>d</i> =0.75	↑ activity level = $\downarrow$ sociability
1982	Fear Duration of orienting Smiling and laughter Soothability	Sociability	sociability Fear vs. sociability	Large: <i>d</i> =0.93 Moderate: <i>d</i> =0.63	<ul> <li>↑ distress to limitations = ↓ sociability</li> <li>↑ fear = ↓ sociability</li> </ul>
Penela et al., 2015	<ul><li>2, 3 years: Behavioral inhibition</li><li>5 years: Emotion regulation</li></ul>	<b>7 years</b> Social competence	Emotion regulation vs. social competence	No significant: $d=0.15$	↑ emotion regulation = ↑ social competence
Van Hecke et al., 2007	<b>15m</b> Social fearfulness Inhibitory control Low pleasure Soothability	<b>2.5 years</b> Social competence	Inhibitory control vs. social competence Low pleasure vs. social competence	Large: <i>d</i> =0.87 Moderate: <i>d</i> =0.52	<ul> <li>↑ inhibitory control = ↑ social competence</li> <li>↑ low pleasure = ↑ social competence</li> </ul>
	Southability		Negative affect (2 years) vs. social cognition	Large: <i>d</i> =-1.32	↑ negative affect (2 years) = $\downarrow$ social cognition
Salley et al., 2013	2, 3 years Surgency/extraversion Negative affect	<b>4 years</b> Social awareness Social cognition	Negative affect (2 years) vs. social-communication	Large: <i>d</i> =-3.53	↑ negative affect (2 years) = $\downarrow$ social- communication
	Effortful control	Social-communication Social motivation	Negative affect (2 years) vs. social motivation	Large: <i>d</i> =-1.86	↑ negative affect (2 years) = $\downarrow$ social motivation
			Negative affect (3 years) vs.	Large: <i>d</i> =-2.08	↑ negative affect (3 years) = $\downarrow$ social

			social cognition		cognition
			Negative affect (3 years) vs. social-communication	Large: <i>d</i> =-1.19	$\uparrow$ negative affect (3 years) = $\downarrow$ social- communication
			Surgency/extraversion (3 years) vs. social motivation	Large: <i>d</i> =-3.37	↑ surgency/extraversion (3 years) = $\downarrow$ social motivation
			Surgency/extraversion (3 years) vs. social-communication	Large: <i>d</i> =-1.16	$\uparrow$ surgency/extraversion (3 years) = $\downarrow$ social-communication
			Effortful control (3 years) vs. social awareness	Large: <i>d</i> =-1.67	↑ effortful control (3 years) = ↓ social awareness
			Temperamental difficulty (8m) vs. social competence (2 years)	Moderate: <i>d</i> =-0.49	↑ temperamental difficult $(8m) = \downarrow$ social competence (2 years)
			Temperamental difficulty (8m) vs. social competence (3 years)	Moderate: $d=-0.47$	↑ temperamental difficult $(8m) = \downarrow$ social competence (3 years)
Houck, 1999	8m, 12m, 2 years Temperamental difficulty	12m, 2, 3 years Social competence	Temperamental difficulty (12m) vs. social competence (2 years)	Moderate: $d=-0.49$	↑ temperamental difficult $(12m) = \downarrow$ social competence (2 years)
	unicuty		Temperamental difficulty (12m) vs. social competence (3 years)	Moderate: $d=-0.63$	↑ temperamental difficult $(12m) = \downarrow$ social competence (3 years)
			Temperamental difficulty (2 years) vs. social competence (3 years)	Large: <i>d</i> =-1.07	↑ temperamental difficult (2 years) = $\downarrow$ social competence (3 years)
Bohlin et al., 2005	<b>13-15m</b> Behavioral inhibition	<b>8 years</b> Social competence	Behavioral inhibition vs. attachment security vs. social competence	Small: <i>d</i> =0.28	<ul> <li>↑ behavioral inhibition = ↓ social competence (in insecure infants)</li> <li>↑ behavioral inhibition = ↑ social competence (in secure infants)</li> </ul>
Hwang et al., 2009	<b>18m-3 years</b> Temperament type (i.e., early, intermediate, difficult)	<b>4-6 years</b> Social-adaptive development	No significance	Small: <i>d</i> =0.32	N/A

			Activity vs. peer-relation problems	No significance: <i>d=</i> 0.14	$\uparrow$ activity = $\downarrow$ peer-relation problems
Abulizi et al., 2017	<b>12m</b> Emotionality Activity Shyness Sociability	<b>5.5 years</b> Peer-relation problems	Activity vs. peer-relation problems (adjusted for sex, premature birth, birth weight and order, family income, social support, and parental covariates)	Moderate: <i>d</i> =0.59	↑ activity = ↓ peer-relation problems (adjusted for covariates)
			Sociability vs. peer-relation problems	No significance: $d=0.14$	$\uparrow$ sociability = $\downarrow$ peer-relation problems
	<b>4-5m, 13-15m, 25-</b> <b>27m</b> Negative emotionality	<b>4-5m, 13-15m, 25-</b> <b>27m</b> Communication development Personal-social development	No significance	No significance: d's=0.03 - 0.07	N/A
<b>Observational Me</b>	easures of ER (n=7)		•		
			Arousal vs. social behavior (5 years; moderated by advanced maternal age)	Small: <i>d</i> =0.34	$\uparrow$ arousal = $\uparrow$ social behavior (5 years; moderated by advanced maternal age)
Bowers et al., 2018	<b>4-5 weeks</b> Arousal Excitability Self-regulation	<b>4, 5, 8 years</b> Social behavior	Excitability vs. social behavior (5 years; moderated by advanced maternal age)	Small: <i>d</i> =0.37	$\uparrow$ excitability = $\uparrow$ social behavior (5 years; moderated by advanced maternal age)
	Stress		Self-regulation vs. social behavior (5 years; moderated by advanced maternal age)	Small: <i>d</i> =0.36	↑ self-regulation = ↓ social behavior (5 years; moderated by advanced maternal age)
Henderson, 2007	<b>2 years</b> Negative emotional reactivity	<b>3 years</b> Social competence	Negative emotional reactivity vs. social competence (interaction with supportive parenting)	Large: <i>d</i> =0.98	$\downarrow$ negative emotional reactivity = $\uparrow$ social competence (interaction with supportive parenting)
Rispoli et al., 2013	2, ~4 (preschool) years	5.5 and 6 (kindergarten) years	Negativity (~4 years) vs. social competence	Small: <i>d</i> =0.20	↑ negativity (~4 years) = ↓ social competence

	Negativity	Social competence			
Belskey et al., 2001	<b>15m</b> Negative emotionality	<b>3 years</b> Social competence	Negative emotionality vs. social competence (moderated by low attentional persistence)	No significance: <i>d</i> =0.06	↑ negative emotionality = ↓ social competence (moderated by low attentional persistence)
NICHD Early Child Care Research Network, 2004	<b>6m:</b> Negative mood, Difficult temperament <b>2, 3 years:</b> Affect dysregulation	4.5 years, kindergarten, grade 1 Social skills (i.e.,	Affect dysregulation (2 years) vs. social skills (maternal report)	Small: $d=0.30$	↑ affect dysregulation (2 years) = $\downarrow$ social skills (maternal report)
		cooperation, assertion, self-control, responsibility)	Affect dysregulation (2, 3 years) vs. social skills (teacher report)	Small: <i>d</i> 's=0.22– 0.24	↑ affect dysregulation (2, 3 years) = $\downarrow$ social skills (teacher report)
Cassel, 2008	6m	8m, 10m, 12m, 15m, 18m	Cry-faces vs. initiating behavioral requests (8m)	Large: <i>d</i> =1.28	↑ cry-faces = ↑ initiating behavioral requests (8m)
	Smiles Cry-faces Neutral affect Socially-directed expressions Affective valence	Initiating joint attention (i.e., anticipatory, reactive,	Cry-faces vs. reactive smiling (8m)	Large: <i>d</i> =3.37	$\uparrow$ cry-faces = $\uparrow$ reactive smiling (8m)
		no smiling) Responding to joint attention	Neutral affect vs. reactive smiling (8m and 12m)	Large: <i>d</i> 's=1.96– 2.41	$\downarrow$ neutral affect = $\uparrow$ reactive smiling (8m and 12m)
		Initiating behavioral requests	Neutral affect vs. no smiling (18m)	Large: <i>d</i> =1.81	$\downarrow$ neutral affect = $\downarrow$ no smiling (18m)
Parlade et al., 2009	<b>6m</b> Still-face smiling	8m, 10m, 12m Initiating joint attention Reactive smiling Anticipatory smiling	Still-face smiling vs. anticipatory smiling (8, 10m)	Large: <i>d</i> 's=1.15– 1.42	↑ still-face smiling = ↑ anticipatory smiling (8, 10m)
		Studies Examini	ng Social Communication → ER	Relationship	
<b>Observational M</b>	easures of Social Comn	nunication (n=4)			
Bedford et al., 2017	<ul> <li>6 years: Emotion recognition</li> <li>7 years: Callous unemotional behaviors</li> </ul>	<b>6m</b> Gaze	Gaze vs. callous unemotional behaviors (moderated by low maternal sensitivity)	Moderate: <i>d</i> =0.50	↓ gaze = ↑ callous unemotional behaviors (moderated by low maternal sensitivity)
Van Hecke et al., 2007	<b>15m</b> Social fearfulness Inhibitory control	<b>12m</b> Initiating joint attention	Responding to joint attention vs. inhibitory control	Moderate: <i>d</i> =0.70 Moderate: <i>d</i> =0.75	↑ responding to joint attention = $\downarrow$ inhibitory control

	Low pleasure Soothability	Responding to joint attention Initiating behavioral requests	Initiating behavioral requests vs. soothability		↑ initiating behavioral requests = ↓ soothability
Parlade et al., 2009	<b>2.5 years</b> Dysregulation Internalizing behaviors Externalizing behaviors	<b>9m, 12m</b> IJA Reactive smiling Anticipatory smiling	No significance	Small: <i>d's</i> =0.02– 0.58	N/A
Thompson et al., 1982	<b>19.5m</b> Activity level Distress to limitations Fear Duration of orienting Smiling and laughter Soothability	<b>12.5m</b> Sociability	Sociability vs. fear	Large: <i>d</i> =0.82	↓ sociability = ↑ fear

# CHAPTER 3: Congruence Between Physiological Arousal and Behavioral Affect During an Emotion Regulation Task in Children at Increased Likelihood for Autism Spectrum Disorder

Although autism spectrum disorder (ASD) is defined by differences in social communication, narrow and restricted interests, and repetitive behavior (American Psychiatric Association, 2013), difficulties in regulating emotions are a common and persistent co-occurring feature (Gomez & Baird, 2005; Zwaigenbaum et al., 2005; Bryson et al., 2007; Clifford et al., 2007; Watson et al., 2007; Garon et al., 2009, 2006). Emotion regulation (ER) refers to the processes responsible for modulating the occurrence, intensity, and valence of emotional responses to environmental stimuli. Depending on the context, ER can take on many forms – it may be automatic or controlled, conscious or unconscious, and intrinsic (i.e., regulating one's own emotions) or extrinsic (i.e., regulating another's emotions; Cole et al., 1994; Thompson, 1994; Gross & Thompson, 2007; Gross & Jazaieri, 2014).

Both parental report and direct observation of children with ASD indicate higher rates of negative emotions and lower rates of positive emotions when compared to neurotypical peers (Ben Shalom et al., 2006; Putnam et al., 2006; Garon et al., 2009; Samson et al., 2013; Hirschler Guttenberg et al., 2015; Macari et al., 2017). For example, when compared to typically developing children on the parent-reported Emotion Behavior Checklist (Izard et al., 1974), children with ASD showed more sadness and fear at 24 months of age (Capps et al., 1993). Similarly, Garon et al. (2016) found that toddlers at increased likelihood of ASD were reported to exhibit higher levels of sadness, fear, and anger, and lower levels of pleasure, soothability, as well as some challenges with executive functioning such as inhibitory control and attention, compared to typically developing toddlers at 12 and 24 months. Direct behavioral observation of ER has shown a similar pattern, such that toddlers at increased likelihood of ASD display elevated rates of fear (Macari et al., 2018) and negative affect (Sacrey et al., 2021) during emotionally salient tasks and lower rates of positive affect during free play (Filliter et al., 2015) compared to typically developing peers. Thus, toddlers at increased likelihood of ASD show continuity in emotional dysregulation from 12 to 24 months of age. These difficulties may forecast the emergence of later internalizing and externalizing behavior, as well as give rise to other mental health disorders commonly co-occurring with ASD during the school-age period

(Leyfer et al., 2006; Gadow et al., 2008; Simonoff et al., 2008; Wood & Gadow, 2010; Salazar et al., 2015).

Although difficulties in ER are well documented in ASD at the behavioral level, much less is understood about how overt emotional expressions are related to internal, physiological states of emotional arousal. Emotions are comprised of multiple response components that unfold over time, a process that requires coordination among cognitive appraisal, a physiological response, and a behavioral response - also referred to as emotional congruence (Hockenbury & Hockenbury, 2010; Hollenstein & Lanteigne, 2014). For example, in a stressful environment, a child may appraise the situation as fear-inducing, thereby eliciting an increase in physiological arousal (increased heart rate and respiration) and a corresponding behavioral response, such as changes in facial affect and/or strategies to mitigate the situation (e.g., running away). The behavioral expression of emotion can be thought of as an external signal, whereas the physiological expression of emotion can be thought of as an internal signal of autonomic nervous system functioning. The autonomic nervous system acts as a regulator by monitoring and adjusting homeostatic functioning in response to internal and external demands. This occurs via the control of the parasympathetic or sympathetic branches (Berntson et al., 1997). Physiological indices of emotional arousal may include cardiac activity, changes in skin sweat levels, and respiration (Kreibig, 2010). In the present study, we aimed to examine the relation between behavioral affect and physiological (heart rate) indices of ER in a cohort of infants at increased likelihood of an ASD diagnosis.

Understanding the congruence between behavioral and physiological response components may provide insight into the degree to which indices are related or synchronized with each other. This may allow for better understanding of individual differences and agerelated changes in response to emotion-eliciting stimuli (Lacey et al., 1963; Davidson, 2001). Moreover, patterns of emotional congruence may also elucidate the mechanisms underlying emotional dysregulation in ASD. Few studies have examined behavioral-physiologic congruence in response to emotion-eliciting stimuli in young children with ASD. Findings have been mixed due to a range of emotions assessed and variation in selected emotion tasks and physiological indices. For example, Zantinge et al. (2019) examined emotional expression and heart rate in response to fear in autistic children and matched controls (age range: 41-81 months). Findings revealed no significant correlation (i.e., congruence) between behavioral affect and heart rate in typical developing children or those with ASD. In contrast, Vernetti et al. (2020) showed that change in skin conductance was positively correlated with emotional expressions during anger and fear challenges in both typically developing toddlers and those with ASD (mean age: 22.7 months). Furthermore, Stein et al. (2014) demonstrated that physiological stress, as measured by skin conductance, was correlated with behavioral distress during dental cleanings in children with ASD (mean age: 8.2 years). These findings highlight the heterogeneity in ER responses in young children with ASD, particularly in behavior-physiologic congruence.

In the present study, we examined congruence between measures of behavioral affect and physiological arousal during an emotion-eliciting task at 6, 12, and 18 months of age in samples of infants at increased likelihood of a diagnosis (IL; younger siblings of children with ASD) and low likelihood (LL; no family history of ASD) for a diagnosis of ASD. Facial affect was coded (i.e., offline from video recordings) by a coder who was blind to group status and heart rate was measured using electrocardiography sensors. Heart rate is a reliable, quantifiable, and readily accessible index of autonomic nervous system function that reflects the combined effects of sympathetic and parasympathetic activity. The sympathetic branch activates the "fight or flight" response, which is reflected by increased heart rate (Levenson, 2014). In contrast, the parasympathetic branch signals the "rest and digest" response, resulting in heart rate deceleration (Selye, 1956; Sapolsky, 2004; Alkon et al., 2014). The balance between both processes promote optimal ER.

Our objectives of this study were to (1) determine whether indices of behavioral affect and heart rate were congruent (i.e., significantly correlated) during positive and negative phases of the ER task, and (2) examine the relation between such congruence and later symptoms of ASD. It was predicted that the relationship between behavioral affect and heart rate will be differentially impacted in IL versus LL infants. Furthermore, behavior-physiologic congruence in the IL group will predict later ASD symptoms. Understanding congruence may help identify changes in internal and external emotional states, the developmental trajectory of ER in young children with ASD, and if congruence is related to overall levels of ASD severity (Zwaigenbaum et al., 2015; Kim et al., 2016). This could facilitate early detection, increased support by caregivers, and tailored interventions to improve ER (Landa, 2008; Dawson & Bernier, 2013).

#### **METHODS**

#### **Participants**

Infant siblings of children with ASD were recruited between 6 and 12 months of age as part of a larger ongoing prospective study of early development in ASD, conducted at three major ASD diagnostic centers in Canada (Glenrose Rehabilitation Hospital in Edmonton, Alberta; Holland Bloorview Kids Rehabilitation Hospital in Toronto, Ontario; IWK Health Centre in Halifax, Nova Scotia). The research ethics boards at each site approved the research study protocol and written informed consent was obtained from the primary caregiver of each participant.

All participants were born between 37 and 42 weeks gestation and had a birth weight greater than 2,500 grams. For IL infant siblings, diagnosis of ASD in the proband (i.e., older sibling) was confirmed by review of clinical records and expert clinical judgment using the DSM-5 (American Psychiatric Association, 2013). None of the IL infants had identifiable neurological disorders or genetic conditions, or any motor or sensory impairment that could potentially account for their ASD presentation. LL control infants were recruited from local communities and had at least one older sibling, but did not have any first- or second-degree relatives with an ASD diagnosis.

Participants were included in this study if they (1) completed the ER task and had both behavioral and physiological data available at 6, 12, and/or 18 months of age, and (2) underwent a 24-month assessment of ASD symptoms.

#### Measures

Behavioral and physiological indices of ER were measured using the ER task at 6, 12, and 18 months. Symptoms of ASD were assessed using the Autism Diagnostic Observation Schedule 2<sup>nd</sup> Edition – Toddler Module (ADOS-2-T) at 24 months by a research-reliable examiner.

## **Emotion Regulation (ER) Task**

The ER Task was adapted from the Laboratory Temperament Assessment Battery (Lab-TAB; Goldsmith & Rothbart, 1996). The Lab-TAB is an observational assessment comprised of activities designed to elicit behavior related to differing dimensions of temperament and emotion (e.g., crying, smiling, changes in facial expression). The Lab-TAB can be used for prelocomotor, locomotor, and preschool ages – both in typical and atypical populations. Moreover, Lab-TAB administration, coding, and scoring are not prescriptive and can be adapted depending on research needs (thus psychometric properties may vary; Gagne et al., 2011; Planalp et al., 2017).

Using the participant guidelines from the mask and toy removal activities in the Lab-TAB manual (Goldsmith & Rothbart, 1996), participants were seated in a highchair at a heightadjustable table with their caregiver seated to the right. All components of the ER task occurred with the child seated in the highchair. Baseline videos were displayed on a computer monitor, which was placed on the table in front of the child, as seen in Figure 1. When the baseline video ended, the computer monitor was placed on the floor next to the examiner, out of the child's sight. Objects and toys used for the positive and negative phases of the ER task were held in a bin next to the examiner – again, out of the child's sight. As shown in Figure 1, our ER task consisted of the following phases:

- (1) Baseline 1 (*neutral*): The child is shown a 120-second video, comprised of 15-second clips of intermixed screensaver images and 'Baby Einstein' clips. The video is accompanied by instrumental music. This neutral activity provides the child an opportunity to acclimatize to the research setting and establish a baseline.
- (2) Bubbles (*positive*): The experimenter blows bubbles towards the child for 90 seconds, while directing the child's attention toward the bubbles.
- (3) Baseline 2 (*neutral*): The child is shown the same two-minute video from Baseline 1. Provides the child an opportunity to return to baseline.
- (4) Toy Play (*positive*): The child is given an attractive toy that lights up and makes noise for 30 seconds.
- (5) Toy Removal (negative): The attractive toy used in Toy Play is removed and placed out of the child's reach (but within sight) for 30 seconds.
- (6) Masks (negative): The experimenter wears a blank mask and sits quietly and still for 15 seconds, followed by wearing a cow mask and sitting quietly and still for an additional 15 seconds.
- (7) Hair Brushing and Face Washing (negative): The experimenter brushes the child's hair with a brush for 15 seconds, followed by gently washing the child's

face (cheeks and forehead) with a baby wipe for an additional 15 seconds.

(8) Baseline 3 (*neutral*): The child is shown the same 120-second video from Baseline 1 and 2. Provides the child an opportunity to return to baseline.

Continuous time-series data were recorded over the 10-minute ER task. Video recordings (for behavioral coding of facial affect) and physiology (heart rate) were time-synched in Noldus Observer XT at time of data collection. Given that the onset and offset of affect intensity were difficult to delineate and facial affect cues may change quickly, interval coding was applied. Specifically, behavioral codes and physiology were collapsed into five-second time intervals across the length of each phase of the ER task. The number of intervals in each phase varied with Baseline phases 1, 2, and 3 having 24 intervals; Bubbles having 18 intervals; Toy play, Toy removal, and Masks phases each having 6 intervals; and hair brushing and face washing phases having 3 intervals each. Mean affect and mean heart rate were also calculated for each phase of the ER task by taking the grand mean of the five-second intervals. For instance, the duration of the Toy Removal phase is 30 seconds and consists of six time intervals (of five seconds each). The mean affect and mean heart rate for Toy Removal was calculated as the sum of the codes/values of the intervals divided by 6.

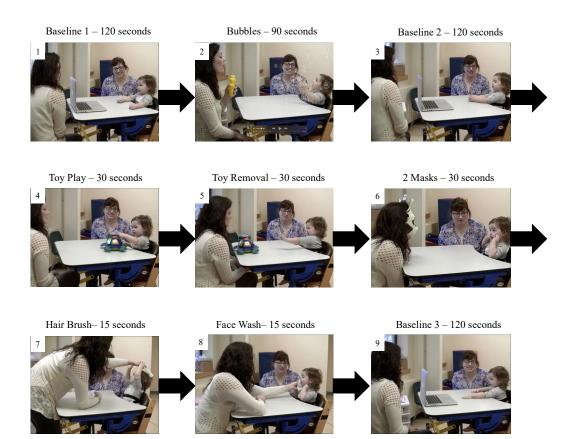


Figure 3.1. Emotion Regulation (ER) Task

*Video Recording*. Children were video-recorded using two video cameras that were controlled digitally by Noldus Observer 13.0 and Media Recorder 4.0 software, all running on a Dell Precision computer (Model 5000 series or above). Observer was used to collect video recordings that were synchronized digitally with the physiological system as described below.

Observational Affect Coding. Behavioral affect was coded off-line from video recordings using Noldus Observer 13 XT behavioral coding software (see Appendix B for coding scheme). Videos were played in real time, and coding for phase (onset and offset) and affect was completed for each participant. Each emotion-eliciting phase was coded continuously across five-second intervals, and codes were mutually exclusive and exhaustive. The child's affective expressions - derived behaviorally from facial and vocal cues - were coded for valence (positive, negative, or neutral) and intensity, which was scored on a 5-point scale (to allow for differentiation of mild displays of affect from extreme displays of affect): +2: Extreme display of positive affect; +1: Mild display of positive affect; 0: Neutral display of affect; -1: Mild display of negative affect; -2: Extreme display of negative affect. Instances in which the child's face was not visible and/or vocal cues were absent were coded as "not codable." Two raters coded 20% of the videos to assess reliability using Cohen's kappa ( $\kappa$ ), with 0.010 – 0.200 representing no to slight agreement, 0.210 - 0.400 representing fair agreement, 0.410 to 0.600 as moderate agreement, 0.610 - 0.800 representing substantial agreement, and 0.810 - 1.000 representing almost perfect agreement (Marston, 2010). When reliability was assessed using a modifier margin of 1 (codes were within + 1 point between raters),  $\kappa = 95\%$ . For gaze,  $\kappa = 89\%$  was achieved when calculating the percentage agreement for duration of gaze codes for the two raters. Both raters were blinded to enrollment group (IL vs. LL) and ASD symptom history, but one rater was involved in study visits at one site.

*Heart rate.* Three pediatric ECG sensors were attached to the child in an inverted triangle, with the right lead placed under the right clavicle, the left lead placed under the left clavicle (both at mid-clavicular line within the rib cage frame), and the ground on the lower left abdomen within the rib cage frame (see Figure 2). Physiological data were acquired using a ProComp Infinity Encoder (T7500M) and Biograph Infinity Software (Version 6). Thought Technology ECG-Flex/pro Sensor Model R4630 was used to collect ECG data, digitized at 2048

The ECG time series that was marked by task onset and offset (previously described in the ER affect coding section) was processed as follows. First, the time series was visually inspected for quality. Records with greater than 5% noise were considered failures. Second, beatto-beat intervals (RR) were extracted from the ECG time series using an adapted version of the Pan-Tompkins algorithm (Pan & Thompkins, 1985; Hamilton & Thompkins, 1986). Values outside of the 1.5\*interquartile envelope were removed. Third, heart rate was computed as the inverse of the RR series (beats per minute, bpm). Heart rate reactivity was calculated for each task by subtracting mean heart rate during Baseline 1 from mean heart rate during each emotioneliciting activity.

To synchronize the video and ECG records, we recorded and digitized a second channel containing a synchronization signal from Noldus Observer 13.0 (Sync Channel). The sync channel contained on and off pulses that occurred when the video recording started and stopped. The signal was sent from Noldus Observer using the computer's COM port to a voltage isolator. The voltage isolator sent the on-off signal to the ProComp Infinity Encoder. Following processing of the physiological data, heart rate and sync signal were imported into Noldus Observer to be synchronized with coded behavior.

Physiological data loss due to movement artifacts, removal of heart rate sensors, or discontinuation of EE Task due to increased levels of negative arousal was 25% at 6 months, 20% at 12 months, and 34% at 18 months. A comparison of data loss by group showed no differences between the three groups at 6 ( $X^2(2) = 1.46$ , p = 0.48), 12 ( $X^2(2) = 2.03$ , p = 0.36), or 18 months ( $X^2(2) = 1.99$ , p = 0.37).

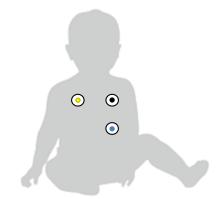


Figure 3.2. Cardiac sensor placement

Hz.

## Autism Diagnostic Observation Schedule – 2<sup>nd</sup> Edition – Toddler Module (ADOS-2-T)

The ADOS-2 (Lord et al., 2012) is a standardized, activity-based assessment designed to elicit communication, social interaction, repetitive behavior, and imaginative play behavior. The ADOS-2 has been demonstrated to reliably distinguish children with ASD from typically developing children and children with intellectual disability (Lord et al., 1989). Subscale scores are derived from a subset of items and summed to generate an overall score. In the present study, the ADOS-2 Toddler module (ADOS-2-T) was administered at 24 months of age to all participants by research-reliable examiners. The Social Affect (SA), Restricted Repetitive Behaviors (RRB), and Total algorithm scores were derived.

#### **Statistical Analyses**

Analyses were run using SPSS Statistics (Version 24, IBM). IL and LL participants who completed at least one ER task at 6, 12, and 18 months were included in the present analyses. Due to non-normal distribution of behavioral and physiological data, non-parametric methods were used across analyses. First, participant demographics were compared between IL and LL groups using Mann-Whitney U for continuous variables and chi-square analyses for categorical variables. Second, as the congruence between behavioral affect and heart rate during the ER task was of primary interest, congruence was measured both continuously (Spearman's rank-order correlations) and categorically (no congruence, negative congruence, positive congruence).

Spearman's rank-order correlations were used to assess congruence using all available interval data. Zero-lagged correlation analyses (i.e., time synced data) were used to identify how changes in one index were concurrently related to changes in the other (Wass et al., 2018). Congruence was analyzed in three ways. First, correlation values between affect and heart rate were calculated across only the negative phases of the ER task as one continuous metric (hereafter referred to as *negative phases*). Second, correlation values between affect and heart rate were calculated across only the positive phases of the ER task as one continuous metric (hereafter, *positive phases*). Third, correlation values between affect and heart rate were also calculated across the baseline phases of the ER task as a continuous metric (hereafter, *baseline*). Following congruence calculations, we were interested in determining whether congruence during the ER task varied between IL and LL infants. Group differences (IL, LL) in congruence

across the *negative phases, positive phases,* and *baseline* at 6, 12, and 18 months were explored using a series of Mann-Whitney U tests.

We were also interested in determining whether the congruence values for each participant was associated with later ADOS-2-T scores. To do this, congruence (i.e., rho) values were categorized as effect sizes to account for both the magnitude and direction of relation between affect and heart rate. As this was an exploratory analysis, this approach would normalize the data and reflect associations akin to effect size estimate interpretations. Rho values (i.e., our measure of congruence) was grouped into three categories: (1) no congruence: rho < +0.10; no relation between affect and heart rate, (2) negative congruence: rho < -0.10; affect and heart rate negatively related), and (3) positive congruence: rho > 0.10; affect and heart rate positively related. Based on the review of rho values, the cutoff +0.10 was chosen as it demarks the value of small effect sizes. Chi-squared analyses were run to assess the association between Group (IL, LL) and Congruence Group (none, negative, positive). Next, a series of Kruskal-Wallis tests examined whether Congruence Group (none, negative, positive) during the negative phases, positive phases, and baseline was associated with later ASD symptoms (24-month ADOS-2-T SA, RRB, Total scores) at 6, 12, and 18 months. Significant Kruskal-Wallis tests were followed up with post hoc tests, using Benjamini and Hochberg corrections (1995) for multiple comparisons.

#### **RESULTS**

#### **Participant Characteristics**

Participant characteristics are summarized in Table 1. Overall, 103 IL (58 boys and 45 girls) and 52 LL (31 boys and 21 girls) children contributed ER data to the present study. There was no significant sex difference by group ( $\chi^2 = 0.154$ , p = 0.414). There were no significant differences between groups for mean affect or mean heart rate during the ER task at 6, 12, and 18 months (p's > 0.05). As expected, there were group differences on the 24-month ADOS-2-T (ASD symptom measure) on both domains: SA (U = 622.500, p = 0.005) and RRB (U = 664.00, p = 0.011), and on the Total score (U = 588.000, p = 0.002).

#### Group Differences in Congruence (Continuous)

Group (IL, LL) differences in congruence across the negative phases, positive phases,

and *baseline* were explored across all time points using Mann-Whitney U tests. Results are presented in Table 2. There were no group differences in congruence across the *negative phases* at 6 (U= 362.000, p = 0.437), 12 (U= 1048.000, p = 0.132), or 18 (U= 358.000, p = 0.973) months. Similarly, there were no group differences in congruence across the *positive phases* at 6 (U= 198.000, p = 0.928), 12 (U= 1030.000, p = 0.054), or 18 (U= 285.000, p = 0.689) months. Finally, there were no group differences in congruence across the *baseline* at 6 (U= 113.000, p = 0.612), 12 (U= 1343.000, p= 0.967), or 18 (U= 151.500, p= 0.427) months.

#### Group Differences in Congruence Category (Categorical)

A series of chi-squared analyses was performed between Group (IL, LL) and Congruence category (no, negative, or positive congruence) during the *negative phases, positive phases,* and *baseline* of the ER task. There were no significant group differences in congruence category across the 6-, 12-, and 18-month time points or across phases (p's > 0.05). Statistical results and details are presented in Table 3.

#### Associations Between Congruence Category and ASD Symptoms for Combined Groups

As there were no significant differences in congruence (measured continuously or categorically) between IL and LL groups, Group was collapsed for the remaining analyses. Multiple Kruskal-Wallis tests were performed with Congruence category as the independent variable and 24-month ADOS-2-T SA, RRB, and Total scores as the dependent variables. Significant associations were followed up by Benjamini and Hochberg corrections to identify Congruence categories that differed from each other. Associations during the *negative phases, positive phases,* and *baseline* of the ER task at 6, 12, 18 months are presented in Table 4. Results described below are for all participants, first for *negative phases,* followed by *positive phases* and *baseline*.

#### 6 months

*Negative phases.* There was a significant association between Congruence category and ADOS-2-T SA (H(2) = 6.590, p = 0.037), RRB (H(2) = 6.520, p = 0.038), and Total score (H(2) = 8.624, p = 0.013). Post hoc analyses (q < 0.05) indicated higher SA (q = 0.017), RRB (q = 0.033), and Total scores (q = 0.050) in the negative congruence category compared to the no

congruence category.

*Positive phases.* There were no significant associations between Congruence category and ADOS-2-T SA (H(2) = 3.147, p = 0.207), RRB (H(2) = 0.624, p = 0.732), or Total score (H(2) = 2.341, p = 0.310) during the positive phases of the ER task.

*Baseline*. There were no significant associations between Congruence category and ADOS-2-T (H(2) = 5.792, p = 0.055), RRB (H(2) = 1.580, p = 0.454), or Total score (H(2) = 5.169, p = 0.075) during the baseline phases of the ER task.

#### 12 months

*Negative phases.* There was a significant association between Congruence category and ADOS-2-T SA (H(2) = 6.463, p = 0.039) and Total scores (H(2) = 6.746, p = 0.034). Post hoc analyses (q < 0.05) indicated higher SA (q = 0.025) and Total score (q = 0.050) in the negative congruence category compared to the no congruence category. In contrast, there was no association between congruence category and ADOS-2-T RRB score (H(2) = 4.277, p = 0.118).

*Positive phases.* There were no significant associations between Congruence category and ADOS-2-T SA (H(2) = 0.403, p = 0.817), RRB (H(2) = 0.125, p = 0.939), or Total scores (H(2) = 0.759, p = 0.684) during the positive phases of the ER task.

*Baseline*. There were no significant associations between Congruence category and ADOS-2-T SA (H(2) = 2.712, p = 0.258), RRB (H(2) = 0.048, p = 0.976), or Total score (H(2) = 1.411, p = 0.494) during the baseline phases of the ER task.

#### 18 months

There were no significant associations between Congruence category and ADOS-2-T SA (p's > 0.05), RRB (p's > 0.05), or Total scores (p's > 0.05) during the *negative*, *positive*, or *baseline* phases of the ER task. Statistical results are presented in Table 3.

#### DISCUSSION

In the present study, we examined behavioral (affect) and physiological (heart rate) congruence during *negative*, *positive*, and *baseline phases* of an ER task at 6, 12, and 18 months in young children at IL and LL for a later diagnosis of ASD. There were two main findings. First, there were no statistically significant differences in congruence between IL and LL groups,

when measured continuously or categorically (i.e., no congruence, negative congruence, or positive congruence). Second, in the combined sample, age-related associations between congruence category and ASD symptoms were seen during the *negative phases* of the ER task, such that participants who were characterized by negative congruence (i.e., increased heart rate and greater negative affect) at 6 and 12 months had higher 24-month ADOS-2-T scores. These findings suggest that behavior-physiologic congruence may not differentiate children who are at an increased versus low likelihood for ASD. However, they demonstrate the complex interaction between behavioral and physiologic indices of ER in young children, and their association with ASD symptoms.

We explored the congruence between affect and heart rate during *positive*, *negative*, and baseline phases of an ER task. Contrary to our prediction, we found no statistically significant differences in congruence between IL and LL groups during any of the ER phases. Our results are similar to those of previous research examining the congruence between facial/bodily expressions of emotion and heart rate in children with and without ASD. Zantinge et al. (2019) did not find group differences in behavior-physiologic congruence during a fear-inducing test protocol in older autistic children compared to neurotypical controls (41-81 months compared to 6-18 months here). Taken together, findings suggest that congruence during negative, positive, and neutral emotion-eliciting conditions is not statistically different between IL and LL groups. This may indicate that young children at IL for ASD have similar levels of behavioral and physiological arousal to typical developing children when their emotions are triggered (by both social and nonsocial stimuli). Although the lack of group differences suggests that congruence itself may not be a meaningful approach for the detection of ASD, it is important to take into account the limited sample size, as well as individual variability. Moreover, it is plausible that examining congruence in phases (positive, negative, and baseline phases), as opposed to in individual tasks (i.e., bubbles, toy play, toy removal, masks, face washing, hair brushing), is influenced by context. For example, Sacrey et al. (2022) demonstrated that affect and heart rate during the toy play, toy removal, masks, face washing, and hair brushing tasks individually differentiated IL and LL children at 6, 12, and 18 months of age - indicating reliability of the task to produce the desired ER response (i.e., positive activities eliciting positive emotions, negative activities eliciting negative emotions). Future work examining behavior-physiologic congruence in a larger sample and during individual tasks is warranted.

It is important to note that congruence is influenced by situational valence and, therefore, largely dependent on the context. For example, in the context of negative stimuli, an increase in negative affect (e.g., frowning or crying) and an increase in physiological arousal (e.g., increase in heart rate) may suggest congruence. Conversely, in the same negative context, an increase in positive affect (e.g., smiling) and increase in physiological arousal (e.g., increase in heart rate) would signal incongruence. Due to the lack of group differences in congruence during the ER task, participant's congruence values were categories into three categories: (1) no congruence (no relation between affect valence and heart rate), (2) negative congruence (affect valence and heart rate positively related), and (3) positive congruence (affect valence and heart rate positively related). This approach was intended to account for both the magnitude of the behavior-physiologic relation, as well as its direction. Similar to the above findings, we found no significant differences in congruence between IL and LL groups.

Although there were no group differences in congruence - measured either continuously or categorically - during the ER task, differences did emerge in the combined sample when examining the association between congruence and ASD symptoms during the negative phases. At both 6 and 12 months, there was a significant association between negative congruence category and ADOS-2-T scores. That is, participants in the negative congruence category had higher SA, RRB, and Total scores than participants in the no congruence category. Together, these findings highlight two key points. First, negative congruence was marked by increased heart rate and increased negative affect (i.e., indices are inversely related). This provides evidence of congruency between behavioral and physiological indices in response to negatively valenced stimuli in a subgroup of toddlers with or without ASD. Second, the association between negative congruence and higher ADOS-2-T scores at 6 and 12 months suggests that children who display increased levels of negative affect also have greater heart rate which are associated with ASD symptoms. This is in accordance with prospective studies of children at increased likelihood for ASD that reported behavioral associations between poor ER and ASD symptom severity (Gomez & Baird, 2005; Zwaigenbaum et al., 2005; Bryson et al., 2007; Clifford et al., 2007; Watson et al., 2007; Garon et al., 2009). For example, parental ratings of temperament on the Infant Behavior Questionnaire-Revised (Gartstein & Rothbart, 2003) revealed lower levels of positive affect, and increased levels of fear, sadness, and anger in IL compared to LL infants, which were associated with subsequent ADOS scores (Garon et al., 2016). Furthermore, using

the Infant Toddler Social-Emotional Assessment (Briggs-Gowan & Carter, 1998), Raza et al. (2019) reported a higher prevalence of concerns related to internalizing (depression, anxiety) and dysregulation (negative emotionality) behavior in 18-month-old IL infants. These behaviors were correlated with severity scores on the ADOS at 36 months. More recently, using the same ER task as the present study, Sacrey et al. (2022) demonstrated an association between facial affect and ASD symptoms in 12- and 18-month-old IL infants, such that affect predicted 24-month Total ADOS-2-T scores. Although some analyses in the present study combine IL and LL infants (unlike the aforementioned studies) and include an index of physiological reactivity, the results provide some support for a relationship between emotional dysregulation and subsequent ASD symptoms, at least in the context of negatively salient tasks. As noted, no associations were observed when analyses were conducted by 'diagnostic likelihood' group, which may be attributable to low power to detect effects in the small sample.

In contrast to the *negative phases*, congruence category was not associated with ADOS-2-T scores at any age for the *positive* and *baseline phases*. This is not surprising given similar behavioral and physiological findings in the ER literature (Sacrey et al., 2021). For example, at a behavioral level, Macari et al. (2018) showed no difference in facial affect between toddlers with or without ASD during *positive*, playful tasks. Likewise, Sacrey et al. (2022) did not find group differences in facial affect between IL and LL infants during the *positive* Toy Play and Bubbles. Furthermore, to our knowledge, only one study to date has examined behavioral-physiologic congruence during *positive* tasks in toddlers with and without ASD. Vernetti et al. (2020) did not find differences in congruence (facial expression correlated with skin conductance) during tasks designed to elicit joy (Bubbles, Puppets, Penguin Toy). Together, in line with the present findings, evidence suggests that IL infants and toddlers may exhibit typical/normative arousal in response to *positive* tasks.

It is also plausible that the tasks designed to elicit positive emotional responses (in our and other studies) did not evoke strong expressions of emotions and/or unable to capture subtle or ambiguous responses to the *positive* stimuli. Similar patterns for *baseline phases* have also been reported. Sacrey et al. (2022) did not find group differences in affect or heart rate between IL and LL infants during *baseline* tasks. These results accord with other physiological studies examining heart rate and heart rate variability during *baseline* tasks in children with and without ASD (Zantinge et al., 2017, 2019; Bazelmans et al., 2019). As baseline tasks are designed to

reflect an individual's capacity to regulate their emotions, as well as their resting state (Appelhans & Luecken, 2006), the lack of significant findings in the literature as well as in the present study was expected.

Limitations to this study must be acknowledged. First, results are derived from a sample than consists of IL and LL siblings and, therefore, may not be generalizable to children with nonfamilial ASD. Second, small sample size and missing physiological and/or behavioral data across the 6-, 12-, and 18-month time points may have limited statistical power to detect differences between IL and LL groups. It is also possible that interval coding (i.e., 5-second intervals) during the ER task, as well as our analytic plan to examine behavior-physiologic congruence (i.e., raw correlation values and effect sizes) may not have accurately captured congruence between the indices. As such, replication of these findings in larger samples and addition methods is warranted. Third, we only assessed time-synced responses - based on the findings from Wass et al. (2018) – however, their sample consisted of neurotypical children. Choosing this approach may have restricted our ability to detect temporal changes between indices due to likelihood status (i.e., IL vs. LL). Fourth, movement artifacts may have reduced the quality of physiological data gathered from these young children, resulting in data exclusion. For example, for participants with extreme reactions to emotion-eliciting stimuli (i.e., hyperreactivity) movement artifacts were often displayed in heart rate data, resulting in failure in quality control. Exclusion of these data may have yielded a sample of participants with muted responses during the ER task. Moreover, whether heart rate alone is a sufficient physiological measure of ER requires further investigation. That is, other physiological indices such as respiratory sinus arrhythmia (Sheinkopf et al., 2019), electrodermal activity (Vernetti et al., 2020), EEG, or EMG may singly or in combination provide a more accurate picture of the ER response and warrant further investigation. General level of arousal prior to stimulation (Lydon et al., 2016) should also be considered. Fifth, given the social nature of the ER task used in this study, use of both social (i.e., interaction with another person) and non-social (e.g., interaction with objects) stimuli is warranted to better elucidate potential confounding effects. That is, whether ER responses are domain-specific (i.e., elicited by nonsocial emotional stimuli) or domain-general (i.e., elicited by both social and nonsocial emotional stimuli) and the impact on behavior and physiological arousal has yet to be ascertained (Vernetti et al., 2020). Finally, we compared congruence category by phases rather than by individual tasks (i.e., bubbles, toy play, toy removal, masks,

face washing, hair brushing). This was chosen because some individual tasks were short in duration and may have been excluded from the analyses. As such, we used phases to maximize data inclusion and comparison across groups and categories.

#### CONCLUSION

Our study compared behavioral-physiologic congruence during an ER task between infants at IL and LL of ASD at 6, 12, and 18 months. No significant differences in congruence between groups were seen; infants at IL and LL of ASD showed similar levels of congruence in response to positive- and negative-emotion-evoking stimuli. It is important to note, that while congruence as defined here did not differentiate IL and LL infants, this does not preclude the possibility that measures of affect and heart rate might separately predict subsequent ASD symptoms or diagnostic status. Indeed, the association between negative congruence and higher ADOS-2-T scores at 6 and 12 months in the combined sample suggests that increased negative affect and greater heart rate was associated with ASD symptoms. Overall, the present study highlights the complexities entailed in examining temporal relations between behavioral and physiological indices of ER during infancy. Future work will investigate how affect and heart rate differentially impact the emergence of ASD.

# Table 3.1. Participant Demographics

Variable	IL Infants Mean (SD)	LL Infants Mean (SD)	All Participants Mean (SD)	Statistic
n	103	52	155	
Sex (boys:girls)	58:45	31:21	89:66	$X^2 = 0.154$ p = 0.414
		6 months		-
n	47	18	65	
Sex (boys:girls)	27:20	9:9	36:29	$X^2 = 0.292$ p = 0.589
Age (mo)	6.453 (0.342)	6.494 (0.322)	6.464 (0.335)	U = 402.000 p = 0.758
Mean Affect	-0.141 (0.191)	-0.042 (0.276)	-0.113 (0.220)	U = 217.000, p = 0.038*
Mean Heart Rate (bpm)	137.303 (9.088)	138.795 (9.688)	137.714 (9.196)	U = 284.000 p = 0.366
Baseline Affect	-0.052 (0.229)	-0.037 (0.297)	-0.048 (0.247)	U = 361.000 p = 0.347
Baseline Heart Rate (bpm)	133.867 (10.597)	136.039 (9.880)	134.469 (10.373)	U = 339.000 p = 0.218
Positive Phase Affect	-0.051 (0.309)	0.031 (0.480)	-0.028 (0.362)	U = 373.000 p = 0.453
Positive Phase Heart Rate (bpm)	264.688 (33.983)	275.895 (22.271)	267.792 (31.422)	U = 318.000 p = 0.124
Negative Phase Affect	-0.957 (1.062)	-0.431 (1.011)	-0.809 (1.067)	U = 292.500 p = 0.069
Negative Phase Heart Rate(bpm)	412.394 (65.266)	422.335 (29.780)	415.147 (65.266)	U = 419.000 p = 0.953
12 months				
п	75	37	112	

Sex (boys:girls)	42:33	22:15	64:48	$X^2 = 0.121$	
Sex (boys.gnis)	72.55		07.70	<i>p</i> = 0.728	
Age (mo)	12.420 (0.474)	12.513 (0.410)	12.450 (0.454)	U = 1119.500 p = 0.097*	
Mean Affect	-0.037 (0.235)	-0.022 (0.192)	-0.033 (0.223)	U = 853.000 p = 0.522	
Mean Heart Rate (bpm)	126.968 (9.263)	127.301 (7.057)	127.062 (8.664)	U = 911.000 p = 0.867	
Baseline Affect	-0.009 (0.300)	-0.121 (0.495)	-0.045 (0.375)	U = 1335.000 p = 0.924	
Baseline Heart Rate (bpm)	125.183 (9.783)	131.304 (13.391)	127.169 (11.390)	U = 1008.000 p = 0.031*	
Positive Phase Affect	0.342 (0.637)	0.124 (0.686)	0.271 (0.658)	U = 1150.500 p = 0.246	
Positive Phase Heart Rate (bpm)	247.906 (22.581)	244.293 (34.394)	246.724 (26.911)	U = 1282.000 p = 0.750	
Negative Phase Affect	-0.587 (1.066)	-0.584 (0.981)	-0.586 (1.034)	U = 1332.000 p = 0.817	
Negative Phase Heart Rate (bpm)	380.866 (46.598)	361.540 (93.769)	374.424 (66.350)	U = 1266.000 p = 0.519	
18 months					
п	40	18	58		
Sex (boys:girls)	20:20	9:9	29:29	$X^2 = 0$ $p = 1.000$	
Age (mo)	18.326 (0.339)	18.149 (0.196)	18.275 (0.314)	U = 207.000 p = 0.040*	
Mean Affect	-0.017 (0.203)	0.027 (0.205)	-0.003 (0.202)	U = 276.000 p = 0.574	
Mean Heart Rate (bpm)	124.147 (11.472)	121.748 (7.733)	123.378 (10.405)	U = 277.000 p = 0.581	
Baseline Affect	-0.017 (0.304)	0.073 (0.277)	0.011 (0.296)	U = 305.500 p = 0.346	

Baseline Heart Rate (bpm)	123.767 (14.225)	122.902 (11.190)	123.499 (13.265)	U = 354.000 p = 0.920	
Positive Phase Affect	0.476 (0.498)	0.486 (0.738)	0.479 (0.576)	U = 353.500 p = 0.913	
Positive Phase Heart Rate (bpm)	237.181 (34.559)	237.562 (15.274)	237.299 (29.779)	U = 344.000 p = 0.788	
Negative Phase Affect	-0.550 (1.144)	-0.277 (0.631)	-0.465 (1.015)	U = 335.000 p = 0.673	
Negative Phase Heart Rate (bpm)	374.004 (40.425)	363.886 (24.218)	370.864 (36.268)	U = 316.000 p = 0.460	
24 months					
ADOS-2-T SA	5.529 (4.766)	2.821 (2.465)	4.862 (4.439)	U = 622.500 p = 0.005**	
ADOS-2-T RRB	2.086 (1.742)	1.143 (1.297)	1.785 (1.680)	U = 664.000 p = 0.011*	
ADOS-2-T Total Severity	7.586 (5.977)	3.964 (3.415)	6.646 (5.575)	U = 588.000 p = 0.002**	

*IL*, infants at increased likelihood of an ASD diagnosis; *LL*, infants at low likelihood of an ASD diagnosis; *ADOS-2-T*, Autism Diagnostic Observation Schedule  $-2^{nd}$  Edition - Toddler module; *SA*, Social Affect; *RRB*, Restricted Repetitive Behavior; *SD*, standard deviation.

\**p* < 0.05, \*\**p* < 0.01

**Table 3.2.** Group differences in behavior-physiological congruence (measured continuously, using Spearman's rank-order correlations) during the positive phases, negative phases, and baseline of the ER paradigm

	IL Infants Mean (SD)	LL Infants Mean (SD)	Mann-Whitney U Test		
	Positive Phases				
6 months	-0.115 (0.296)	-0.143 (0.350)	U = 198.000 p = 0.928		
12 months	0.021 (0.226)	-0.096 (0.305)	U = 1030.000 p = 0.054		
18 months	-0.010 (0.312)	0.017 (0.306)	U = 285.000 p = 0.689		
	Negativ	e Phases			
6 months	-0.270 (0.311)	-0.196 (0.341)	U = 362.000 p = 0.437		
12 months	-0.178 (0.348)	-0.270 (0.307)	U = 1048.000 p = 0.132		
18 months	-0.093 (0.330)	-0.055 (0.314)	U = 358.000 p = 0.973		
	Baseline				
6 months	-0.034 (0.325)	-0.114 (0.392)	U = 113.000 p = 0.612		
12 months	-0.040 (0.267)	-0.057 (0.258)	U = 1343.000 p = 0.967		
18 months	-0.077 (0.286)	-0.016 (0.363)	U = 151.500 p = 0.427		

IL, infants at increased likelihood of an ASD diagnosis; LL, infants at low likelihood of an ASD diagnosis; SD, standard deviation.

**Table 3.3.** Group differences in congruence group (no congruence, negative congruence, positive congruence) during the positive phases, negative phases, and baseline of the ER paradigm

	6 months	12 months	18 months
Positive Phases	$X^2 = 2.160$	$X^2 = 3.260$	$X^2 = 2.315$
	p = 0.340	<i>p</i> = 0.196	p = 0.314
Negative Phases	$X^2 = 1.844$	$X^2 = 4.722$	$X^2 = 0.085$
	p = 0.398	p = 0.094	p = 0.958
Baseline	$X^2 = 0.958$	$X^2 = 1.381$	$X^2 = 0.785$
	<i>p</i> = 0.619	p = 0.501	p = 0.675

	Positive Phases	Negative Phases	Baseline	Benjamini and Hochberg Correction	
		6 months			
ADOS-2-T SA	H = 3.147	H = 6.590	H = 5.792	<i>Negative congruence &gt;</i>	
11005-2-1 51	P = 0.207	<i>p</i> = 0.037*	p = 0.055	No congruence	
ADOS-2-T RRB	H = 0.624	H = 6.520	H = 1.580	<i>Negative congruence &gt;</i>	
ADOS-2-1 KKB	p = 0.732	<i>p</i> = 0.038*	p = 0.454	No congruence	
ADOS-2-T Total	H = 2.341	H = 8.624	H = 5.169	<i>Negative congruence &gt;</i>	
Severity	p = 0.310	<i>p</i> = 0.013*	p = 0.075	No congruence	
	12 months				
	H = 0.403	H = 6.463	H = 2.712	<i>Negative congruence &gt;</i>	
ADOS-2-T SA	p = 0.817	<i>p</i> = 0.039*	p = 0.258	No congruence	
	H = 0.125	H = 4.277	H = 0.048	NS	
ADOS-2-T RRB	p = 0.939	<i>p</i> = 0.118	<i>p</i> = 0.976	143	
ADOS-2-T Total	H = 0.759	H = 6.746	H = 1.411	<i>Negative congruence &gt;</i>	
Severity	p = 0.684	<i>p</i> = 0.034*	<i>p</i> = 0.494	No congruence	
18 months					
ADOS-2-T SA	H = 1.035	H = 1.451	H = 0.672	NS	
	p = 0.596	p = 0.484	p = 0.715	145	
ADOS-2-T RRB	H = 1.283	H = 0.004	H = 5.302	NS	
ADU5-2-1 KKD	p = 0.527	<i>p</i> = 0.998	<i>p</i> = 0.071	145	
ADOS-2-T Total	H = 0.531	H = 0.821	H = 0.009	NS	
Severity	p = 0.767	<i>p</i> = 0.663	<i>p</i> = 0.995	140	

**Table 3.4.** Associations between congruence group (no congruence, negative congruence, positive congruence) and ASD symptoms for IL and LL participants combined during the positive phases, negative phases, and baseline of the ER paradigm

ADOS-2-T, Autism Diagnostic Observation Schedule  $-2^{nd}$  Edition - Toddler module; SA, Social Affect; RRB, Restricted Repetitive Behavior.

## CHAPTER 4: The Role of Emotion Regulation and Joint Attention in the Emergence of Autism Spectrum Disorder: An Infant Siblings Study

Autism spectrum disorder (ASD) is characterized by differences in social communication, as well as the presence of restricted, repetitive behavior and interests (American Psychiatric Association, 2013). Albeit not a core symptom, difficulties with emotional regulation constitute a persistent, co-occurring feature of ASD. Emotion regulation (ER) refers to the processes responsible for modulating the occurrence, valence, and intensity of emotional reactions to external stimuli. Depending on the context, ER may take on multiple forms; conscious or unconscious, automatic or controlled, and/or intrinsic or extrinsic (Cole et al., 1994; Thompson, 1994; Gross & Thompson, 2007; Gross & Jazaieri, 2014). The development of effective ER strategies helps to facilitate the growth of foundational social skills and communicative abilities. That is, ER undergirds a child's behavioral and physiological flexibility, which is fundamental to maintaining an optimal level of arousal necessary for social and cognitive engagement (Laurent & Rubin, 2004). For example, when in an optimal state of arousal, a child can successfully navigate and attend to aspects of their social environment, enhancing their ability to initiate and maintain social interactions, effectively communicate, and adapt to changes in their environment (Williamson & Anzalone, 2001). Given that ER is fundamentally linked to the development of later social skills (Eisenberg et al., 2000; Eisenberg et al., 2010), it is no surprise that emotional dysregulation, as seen in ASD, predicts later differences in social communication, affective expression, and reciprocal play (DeGangi & Breinbauer, 1997). In fact, a high degree of concordance has been shown between emotional dysregulation and the core features of ASD (Maskey et al. 2013; Totsika et al. 2011).

In addition to ER difficulties, differences in joint attention are a salient and defining feature of ASD (Dawson et al., 2004). Defined as the ability to coordinate visual attention with a person, and subsequently shift gaze towards an object or event (Mundy & Gomez, 1998), joint attention is a significant predictor of social competence and cognitive outcomes in typically developing children and those with neurodevelopmental disorders, including ASD. For example, previous studies of children diagnosed with ASD report fewer instances of joint attention when compared to neurotypical peers or children with developmental delays (Mundy et al., 1986, 1990). Similarly, studies of children at increased likelihood of an ASD diagnosis (IL; younger

siblings of children diagnosed with ASD) also show lower levels of social communicative behavior, including initiating joint attention (IJA, e.g., looking from a toy to a play partner to invite them into the play) and responding to bids for joint attention (RJA, e.g., looking to where a play partner points), than siblings of children with no familial risk for ASD. These differences in RJA and IJA are associated with later ASD symptomatology (Cassel et al. 2007; Goldberg et al. 2005; Presmanes et al. 2007; Rozga et al. 2011; Yirmiya et al. 2006). Because ER influences several processes that are fundamental for the successful engagement in joint attention, for example, modulating eye contact, gestures, and attention (Gulsrud et al., 2010), an examination of the interplay between ER and joint attention and their role in the emergence of ASD is warranted.

In the present study, we examined the relation between early ER, joint attention, and subsequent ASD emergence in a sample of infants at IL and low likelihood (LL; no family history of ASD) for a diagnosis of ASD. Behavioral and physiological indices of ER were assessed during an emotion-eliciting task at 12 months of age, followed by joint attention at 18 months using a modified version of the Early Social Communication Scales. At 24 months, all participants underwent an assessment to measure ASD symptoms. In accordance with findings from typical developing populations, where effective ER is predictive of enhanced social communication and functional outcomes (Gross, 2002; Gross & John, 2003; Laurent & Rubin, 2004), we predicted that (1) infants in the IL group would display atypical ER and lower rates of joint attention compared to LL infants, (2) ER and joint attention would predict subsequent ASD symptoms at 24 months and, (3) joint attention would mediate the relationship between ER and later ASD symptoms. Elucidating this fundamental relation may provide insights into the early emotional and social-communicative impairments characterized by ASD.

#### **METHODS**

#### **Participants**

Infant siblings of children with ASD were recruited between 6 and 12 months of age as part of a larger ongoing prospective study of early development in ASD, conducted at three major ASD diagnostic centers in Canada (Glenrose Rehabilitation Hospital in Edmonton, Alberta; Holland Bloorview Kids Rehabilitation Hospital in Toronto, Ontario; IWK Health Centre in Halifax, Nova Scotia). The research ethics boards at each site approved the research protocol, and written informed consent was obtained from the primary caregiver of each participant.

Participants were born between 36 and 42 weeks gestation and had a birth weight greater than 2500 grams. For IL infant siblings, diagnosis of ASD in the proband (i.e., older sibling) was confirmed by review of clinical records and expert clinical judgment using the DSM-5 (American Psychiatric Association, 2013). None of the IL infants had identifiable neurological disorders or genetic conditions, or any motor or sensory impairment that could potentially account for their ASD presentation. LL infants were recruited from local communities and had at least one older sibling, but did not have any first- or second-degree relatives with an ASD diagnosis.

Participants were included in this study if they (1) completed the ER task with behavioral and physiological data available at 12 months, (2) completed the modified version of the Early Social Communications Scales (mESCS) at 18 months, and (3) underwent an 18 or 24-month assessment of ASD symptoms.

## Measures

Behavioral and physiological indices of ER were measured using the ER task at 12 months. Joint attention was measured at 18 months using the mESCS. Symptoms of ASD were assessed using the Autism Diagnostic Observation Schedule  $-2^{nd}$  Edition - Toddler Module (ADOS-2-T) at 18 and/or 24 months by an expert clinician.

#### **Emotion Regulation (ER) Task**

The ER task was adapted from the Laboratory Temperament Assessment Battery (Lab-TAB; Goldsmith & Rothbart, 1996). The Lab-TAB is an observational assessment comprised of activities designed to elicit behavior related to differing dimensions of temperament and emotion (e.g., crying, smiling, changes in facial expression, etc.). The Lab-TAB can be used for prelocomotor, locomotor, and preschool ages – both in typical and atypical populations. Moreover, Lab-TAB administration, coding, and scoring are not prescriptive and can be adapted depending on research needs (thus psychometric properties may vary; Gagne et al., 2011; Planalp et al., 2017). Using the participant guidelines from the mask and toy removal activities in the Lab-TAB manual (Goldsmith & Rothbart, 1996), participants were seated in a highchair at a heightadjustable table with their caregiver seated to the right. All components of the ER task occurred with the child seated in the highchair. Baseline videos were displayed on a computer monitor, which was placed on the table in front of the child, as seen in Figure 1. When the baseline video ended, the computer monitor was placed on the floor next to the examiner, out of the child's sight. Objects and toys used for the positive and negative phases of the ER task were held in a bin next to the examiner – again, out of the child's sight. As shown in Figure 1, our ER task consisted of the following phases:

- (1) Baseline 1 (*neutral*): The child is shown a 120-second video, comprised of 15-second clips of intermixed screensaver images and 'Baby Einstein' clips. The video is accompanied by instrumental music. This neutral activity provides the child an opportunity to acclimatize to the research setting and establish a baseline.
- (2) Bubbles (*positive*): The experimenter blows bubbles towards the child for 90 seconds, while directing the child's attention toward the bubbles.
- (3) Baseline 2 (*neutral*): The child is shown the same two-minute video from Baseline 1. Provides the child an opportunity to return to baseline.
- (4) Toy Play (*positive*): The child is given an attractive toy that lights up and makes noise for 30 seconds.
- (5) Toy Removal *(negative)*: The attractive toy used in Toy Play is removed and placed out of the child's reach (but within sight) for 30 seconds.
- (6) Masks (negative): The experimenter wears a blank mask and sits quietly and still for 15 seconds, followed by wearing a cow mask and sitting quietly and still for an additional 15 seconds.
- (7) Hair Brushing and Face Washing *(negative)*: The experimenter brushes the child's hair with a brush for 15 seconds, followed by gently washing the child's face (cheeks and forehead) with a baby wipe for an additional 15 seconds.
- (8) Baseline 3 (neutral): The child is shown the same 120-second video from Baseline 1 and 2. Provides the child an opportunity to return to baseline.

Continuous time-series data were recorded over the 10-minute ER task. Video recordings

(for behavioral coding of facial affect) and physiology (heart rate) were time-synched in Noldus Observer XT at time of data collection. Given that the onset and offset of affect intensity were difficult to delineate and facial affect cues may change quickly, interval coding was applied. Specifically, behavioral codes and physiology were collapsed into five-second time intervals across the length of each phase of the ER task. The number of intervals in each phase varied with Baseline phases 1, 2, and 3 having 24 intervals; Bubbles having 18 intervals; Toy play, Toy removal, and Masks phases each having 6 intervals; and hair brushing and face washing phases having 3 intervals each. Mean affect and mean heart rate were also calculated for each phase of the ER task by taking the grand mean of the five-second intervals. For instance, the duration of the Toy Removal phase is 30 seconds and consists of six time intervals (of five seconds each). The mean affect and mean heart rate for Toy Removal was calculated as the sum of the codes/values of the intervals divided by 6.

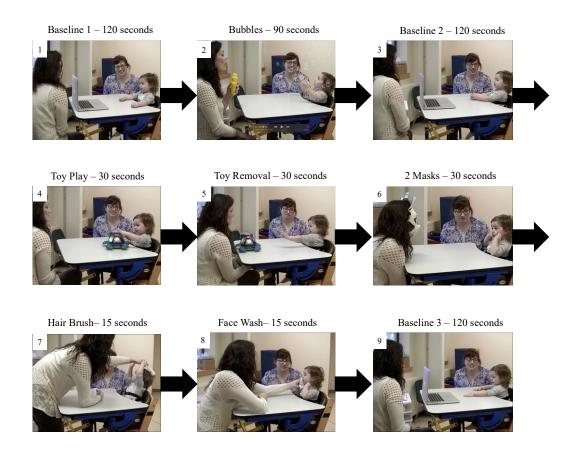


Figure 4.1. Emotion Regulation (ER) Task

*Video Recording.* Children were video-recorded using two video cameras that were controlled digitally by Noldus Observer 13.0 and Media Recorder 4.0 software, all running on a Dell Precision computer (Model 5000 series or above). Observer was used to collect video recordings that were synchronized digitally with the physiological system as described below.

Observational Affect Coding. Behavioral affect was coded off-line from video recordings using Noldus Observer 13 XT behavioral coding software (see Appendix B for coding scheme). Videos were played in real time, and coding for phase (onset and offset) and affect was completed for each participant. Each emotion-eliciting phase was coded continuously across five-second intervals, and codes were mutually exclusive and exhaustive. The child's affective expressions – derived behaviorally from facial and vocal cues – were coded for *valence* (positive, negative, or neutral) and intensity, which was scored on a 5-point scale (to allow for differentiation of mild displays of affect from extreme displays of affect): +2: Extreme display of positive affect; +1: Mild display of positive affect; 0: Neutral display of affect; -1: Mild display of negative affect; -2: Extreme display of negative affect. Instances in which the child's face was not visible and/or vocal cues were absent were coded as "not codable." Two raters coded 20% of the videos to assess reliability using Cohen's kappa ( $\kappa$ ), with 0.010 – 0.200 representing no to slight agreement, 0.210 - 0.400 representing fair agreement, 0.410 to 0.600 as moderate agreement, 0.610 - 0.800 representing substantial agreement, and 0.810 - 1.000 representing almost perfect agreement (Marston, 2010). When reliability was assessed using a modifier margin of 1 (codes were within + 1 point between raters),  $\kappa = 95\%$ . For gaze,  $\kappa = 89\%$  was achieved when calculating the percentage agreement for duration of gaze codes for the two raters. Both raters were blinded to enrollment group (IL vs. LL) and ASD symptom history, but one rater was involved in study visits at one site.

*Heart rate.* Three pediatric ECG sensors were attached to the child in an inverted triangle, with the right lead placed under the right clavicle, the left lead placed under the left clavicle (both at mid-clavicular line within the rib cage frame), and the ground on the lower left abdomen within the rib cage frame (see Figure 2). Physiological data were acquired using a ProComp Infinity Encoder (T7500M) and Biograph Infinity Software (Version 6). Thought Technology ECG-Flex/pro Sensor Model R4630 was used to collect ECG data, digitized at 2048 Hz.

The ECG time series that was marked by task onset and offset (previously described in

the ER affect coding section) was processed as follows. First, the time series was visually inspected for quality. Records with greater than 5% noise were considered failures. Second, beat-to-beat intervals (RR) were extracted from the ECG time series using an adapted version of the Pan-Tompkins algorithm (Pan & Thompkins, 1985; Hamilton & Thompkins, 1986). Values outside of the 1.5\*interquartile envelope were removed. Third, heart rate was computed as the inverse of the RR series (beats per minute, bpm). Heart rate reactivity was calculated for each task by subtracting mean heart rate during Baseline 1 from mean heart rate during each emotion-eliciting activity.

To synchronize the video and ECG records, we recorded and digitized a second channel containing a synchronization signal from Noldus Observer 13.0 (Sync Channel). The sync channel contained on and off pulses that occurred when the video recording started and stopped. The signal was sent from Noldus Observer using the computer's COM port to a voltage isolator. The voltage isolator sent the on-off signal to the ProComp Infinity Encoder. Following processing of the physiological data, heart rate and sync signal were imported into Noldus Observer to be synchronized with coded behavior. Physiological data loss due to movement artifacts, removal of heart rate sensors, or discontinuation of EE Task due to increased levels of negative arousal was 20% at 12 months.

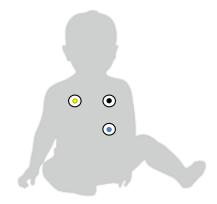


Figure 4.2. Cardiac sensor placement

#### Modified Version of the Early Social Communications Scales (mESCS)

A modified version of the Early Social Communication Scales (mESCS) was used at 18 months of age (Mundy et al., 2003). Using standardized presses with toys and gestures, the mESCS is a structured assessment that examines an infant's ability to initiate and respond to social requests and joint attention bids with an examiner (i.e., non-verbal social communication).

The examiner and child were seated facing each other across a height-adjustable table. The child was seated in the caregiver's lap or in a highchair. The session was video-recorded using one or two video cameras that were controlled digitally using Noldus Observer 13.0 and Media Recorder 4.0 software, all running on a Dell Precision computer (Model 5000 series and above). It was ensured that the video camera was positioned to capture a three-quarter to fullface view of the child, while also capturing the profile view of the tester. The objects used during the assessment were placed on a small table within the child's view, but out of his/her reach. Three windup toys and three hand-held mechanical toys (party noise maker, squeeze toy, bellows toy) were presented across a total of 18 presses (i.e., three consecutive presses per toy). A book with large distinct pictures was also presented across 6 presses.

The experimenter-child interactions were coded off-line from video recordings and yielded scores for joint attention. The frequency by which a child pointed to a toy, initiated eye contact while manipulating a toy, and alternated eye gaze between a toy and the examiner was coded as initiating joint attention (IJA). The frequency by which a child correctly turned his/her head and eyes in response to the examiner's points during the book task was coded as responding joint attention (RJA). Two coders (SR and LRS) independently coded the data from 75 videos and the intra-class correlation estimate of their reliability was 0.932.

## Autism Diagnostic Observation Schedule – 2<sup>nd</sup> Edition – Toddler Module (ADOS-2-T)

The ADOS-2 (Lord et al., 2012) is a standardized, activity-based assessment designed to elicit communication, social interaction, repetitive behavior, and imaginative play behavior. The ADOS-2 has been demonstrated to reliably distinguish children with ASD from typical developing children and children with intellectual disability (Lord et al., 1989). In the present study, the ADOS-2 Toddler module was administered at 18 and 24 months of age by research-reliable examiners. Calibrated Severity Scores (CSS) were calculated. CSS is a standardized metric used to assess ASD symptoms as a clinical entity distinct from adaptive and cognitive

differences (Gotham et al., 2009). It allows for comparability across modules and scores range between 1 to 10, where 1–3 indicates no ASD, 4–5 indicates spectrum, and 6–10 indicates ASD. As a subset of children were unable to complete in-person visits due to COVID-19 restrictions, 18- and/or 24-month ADOS-2-T CSS scores were used, where available. Previous longitudinal analyses of CSS have indicated stability of CSS from 12 to 24 months of age in children with ASD and, thus, a valid indicator of autism severity (Shumway et al., 2012). To confirm this, a series of Mann-Whitney U analyses comparing all variables in the present study (i.e., heart rate, affect, IJA, RJA, ADOS-2-T CSS) by age of the ADOS-2-T (18 or 24 months) revealed no significant differences (p's > 0.12). As such, 18- (n = 48) and 24-month (n = 135) ADOS-2-T CSS scores were combined.

## **Statistical Analyses**

Analyses were run using SPSS Statistics (Version 24, IBM) and macro-program PROCESS 3.4 (Hayes, 2013). IL and LL participants who completed the ER task at 12 months, the mESCS at 18 months, and the ADOS-2-T at 18 or 24 months were included in the present analyses. Due to non-normal distribution of behavioral and physiological data, non-parametric methods were used across analyses.

First, participant demographics were compared between IL-ASD (i.e., infants at increased likelihood for ASD who meet classification criteria for ASD using the ADOS-2-T), IL-non ASD (i.e., infants at increased likelihood for ASD who do not meet classification criteria for ASD using the ADOS-2-T), and LL (i.e., toddlers who did not have first- or second-degree relatives with an ASD diagnosis and do not meet classification criteria for ASD using the ADOS-2-T) groups using Kruskal Wallis tests for continuous variables and chi-square analyses for categorical variables. Second, the mean affect and mean heart rate were calculated for the positive and negative activities of the ER task, by taking the overall mean of the respective activities. For example, the mean affect and mean heart rate during the negative activities (i.e., toy removal, masks, hair brushing and face washing) was calculated (hereafter referred to as *negative phases*). Similarly, the mean affect and mean heart rate during the positive activities (i.e., bubbles and toy play) was calculated (hereafter referred to as *positive phases*).

The primary goal was to investigate the relation between ER (heart rate and affect) and subsequent ADOS-2-T CSS, using parallel mediation with moderation analysis. We were

interested in the mediating effects of IJA and RJA, as well as the moderating effect of Group (IL-ASD, IL-non ASD, LL) on this relation, which were treated as variables in the regression equations. First, parallel mediation analysis was used to identify and elucidate the relation between the independent variable (heart rate and affect) and the dependent variable (ADOS-2-T CSS), which may have been affected by the interaction with IJA and/or RJA. Because heart rate and affect show poor congruence (Raza et al., 2022), they were considered in separate analyses. This would explain "why" heart rate and/or affect and ADOS-2-T CSS are related. Alternatively, moderation analysis was used to identify the Group (IL-ASD, IL-non ASD, LL) conditions under which the independent variable (heart rate and affect) is related to the dependent variable (ADOS-2-T CSS). That is, moderation would explain "when" heart rate and/or affect and ADOS-2-T CSS are related. In sum, PROCESS was operated using two independent variables (heart rate and affect), two mediators (IJA and RJA), one multicategorical moderator (Group: IL-ASD, IL-non ASD, LL), and one dependent variable (ADOS-2-T CSS). Models for heart rate and affect were run separately, as well as for *negative phases* and *positive phases*. We used 5,000 bootstrap samples and determined the mediating and moderating effect within 95% confidence intervals.

#### RESULTS

## **Participant Characteristics**

Participant characteristics are summarized in Table 1. Overall, 27 IL-ASD (21 boys and 6 girls) 90 IL-non ASD (42 boys and 48 girls), and 54 LL (35 boys and 19 girls) contributed ER data to the present study. There was a significant sex difference by group ( $\chi^2 = 10.034$ , p = 0.007). There were no significant differences between group for mean heart rate during the positive phases (H = 1.717, p = 0.424), mean affect during the positive phases (H = 3.140, p = 0.208), mean affect during the negative phases (H = 5.113, p = 0.078), and IJA (H = 5.572, p = 0.062). There were significant differences by group for mean heart rate during the negative phases (H = 8.691, p = 0.013) and RJA (H = 11.368, p = 0.003). As expected, there was a group difference on the ADOS-2-T CSS (H = 56.626, p < 0.001).

#### **Parallel Mediation Analyses with Moderation**

#### **Negative Phases**

Figure 4.3 shows the results of the analysis with RJA  $(M_1)$  and IJA  $(M_2)$  as mediators, Group (W) as the moderator, and Sex as the covariate (as it differed by Group) of the relation between heart rate (X) and ADOS-2-T CSS (Y) during the negative phases. Figure 4.4 shows the results of the analysis with RJA  $(M_1)$  and IJA  $(M_2)$  as mediators, Group (W) as the moderator, and Sex as the covariate of the relation between affect (X) and ADOS-2-T CSS (Y) during the negative phases.

#### ER and Autism Severity

*Heart Rate.* There was no direct relation between heart rate and ADOS-2-T CSS ( $\beta = 0.278$ , p = 0.0520), but there was a moderating effect of Group such that the relation between heart rate and ADOS-2-T CSS ( $\beta = -0.326$ , p = 0.027) was significantly different for the IL-ASD and LL group. There was also an overall Sex effect ( $\beta = 1.008$ , p = 0.011), where boys had higher ADOS-2-T CSS compared to girls.

Affect. There was no direct relation between affect and ADOS-2-T CSS ( $\beta = -1.591$ , p = 0.204), nor any moderating effects of group (p's > .05). There was a Sex effect ( $\beta = 0.891$ , p = 0.019), suggesting that boys had higher ADOS-2-T CSS compared to girls.

#### ER and Joint Attention

*Heart rate*. Heart rate was inversely related to RJA ( $\beta = -4.283$ , p = 0.047) and IJA ( $\beta = -2.094$ , p = 0.005), suggesting that increased heart rate during the *negative tasks* was associated with decreased levels of IJA and RJA. Furthermore, moderation revealed significant interactions between heart rate and Group for RJA (IL-ASD versus LL,  $\beta = 4.427$ , p = 0.047) and IJA (IL-ASD vs. IL-non ASD,  $\beta = 2.260$ , p = 0.031; IL-ASD versus LL,  $\beta = 2.010$ , p = 0.009), such that the effect of heart rate on RJA and IJA was more pronounced in IL-ASD infants.

Affect. Affect was positively related to RJA ( $\beta = 39.797$ , p = 0.049) and IJA ( $\beta = 16.862$ , p = 0.033), suggesting that lower levels of negative affect during the negative tasks was associated with increased levels of IJA and RJA. The relation between affect and IJA was moderated by Group ( $\beta = -19.073$ , p = 0.044), with the relation inversely related in IL-ASD infants compared to IL-non ASD infants.

#### Joint Attention and Autism Severity

*Heart rate*. RJA ( $\beta$  = -0.014, p = 0.080) and IJA ( $\beta$  = -0.029, p = 0.197) were not associated with ADOS-2-T CSS.

Affect. RJA ( $\beta$  = -0.019, p = 0.007), but not IJA ( $\beta$  = -0.027, p = 0.125), was associated with ADOS-2-T CSS. There was an inverse relations between RJA and ADOS-2-T CSS, suggesting that lower levels of RJA were associated with higher scores on the ADOS-2-T CSS.

#### **Positive Phases**

Figure 4.5 shows the results of the analysis with RJA  $(M_1)$  and IJA  $(M_2)$  as mediators, Group (W) as the moderator, and Sex as a covariate (as it differed by Group) of the relation between heart rate (X) and ADOS-2-T CSS (Y) during the positive phases. Figure 4.6 shows the results of the analysis with RJA  $(M_1)$  and IJA  $(M_2)$  as mediators, Group (W) as the moderator, and Sex as a covariate of the relation between affect (X) and ADOS-2-T CSS (Y) during the positive phases.

#### ER and Autism Severity

*Heart Rate.* There was no direct effect of heart rate ( $\beta = -0.100$ , p = 0.222) on ADOS-2-T CSS, but there was a moderating effect of Group, with this relation being significant in the IL-ASD group (compared to the IL non-ASD ( $\beta = -3.145$ , p < 0.001) and LL ( $\beta = -3.556$ , p < 0.001) groups). There was also a Sex ( $\beta = 1.079$ , p = 0.005) effect; boys had higher ADOS-2-T CSS compared to girls.

Affect. Although there was no direct ( $\beta = -2.525$ , p = 0.138) effect of affect on ADOS-2-T CSS, there was a moderating effect of Group, with this relation being significant in the IL-ASD group (compared to the IL non-ASD ( $\beta = -3.738$ , p < 0.001) and LL ( $\beta = -4.199$ , p < 0.001) groups). There was also a Sex ( $\beta = 0.861$ , p = 0.016) effect, suggesting that boys have higher ADOS-2-T CSS compared to girls.

#### **ER** and Joint Attention

*Heart rate*. Heart rate is unrelated to RJA ( $\beta = 1.105$ , p = 0.432) and IJA ( $\beta = 0.281$ , p = 0.572) at 18 months. There was no effect of moderation by Group on RJA nor IJA (p's > 0.05).

Affect. Affect was positively related to RJA ( $\beta = 57.310$ , p = 0.046), but not IJA ( $\beta = 0.812$ , p = 0.943), at 18 months, suggesting that decreased displays of negative affect during the positive tasks was associated with increased levels of RJA.

#### Joint Attention and Autism Severity

*Heart rate*. RJA ( $\beta$  = -0.025, *p* = 0.001) and IJA ( $\beta$  = -0.044, *p* = 0.034) were negatively associated with ADOS-2-T CSS; that is, reduced levels of RJA and IJA were related to higher ADOS-2-T CSS.

Affect. RJA ( $\beta$  = -0.018, p = 0.009) and IJA ( $\beta$  = -0.033, p = 0.046) were negatively associated with ADOS-2-T CSS; that is, reduced levels of RJA and IJA were related to higher ADOS-2-T CSS.

#### DISCUSSION

In the present study, we examined the role of early emotion regulation (ER), as measured by behavioral (affect) and physiological (heart rate) indices, and joint attention in the emergence of ASD symptoms in young children at IL and LL for a diagnosis of ASD. There were three main findings. First, although there were no direct associations between ER and ASD symptoms, there was a moderating effect of Group, with the relation being significant for the IL-ASD infants compared to IL-non ASD and LL infants. Second, 12-month ER and 18-month joint attention were directly associated across all participants. That is, during the negative phases, increased heart rate and displays of negative affect were associated with reduced levels of joint attention. This association was further moderated by the IL-ASD group. Conversely, during the positive phases, lower levels of negative affect were associated with higher levels of joint attention. Third, joint attention during the negative and positive phases showed inverse relations with ASD symptoms across all participants, suggesting that lower levels of joint attention were associated with higher levels of ASD symptoms. These results inform the understanding of developmental pathways between early ER, joint attention, and ASD emergence in young children and suggest a complex interplay between ER and joint attention in children who are at an increased likelihood for ASD.

There were no direct associations between 12-month measures of ER (during the negative and positive phases) and 24-month ADOS-2-T CSS. Yet, there was evidence that the relation

varied by group and sex. During the negative phases, heart rate was associated with higher ADOS-2-T CSS for participants who were in the IL-ASD group compared to IL-non ASD and LL groups, as well as in boys compared to girls. Similar findings were observed during the positive phases of the ER task, where affect was also associated with higher ADOS-2-T CSS in the IL-ASD group, and among boys. The moderating effects of group and sex are consistent with previous literature. That boys had higher ADOS scores compared to girls is in alignment with epidemiological studies that report a 4.2 times prevalence of ASD among boys compared to girls (Maenner et al., 2021). Moreover, group differences in the relation between early ER and subsequent ASD symptoms is in line with the infant sibling literature that report behavioral associations between poor ER/temperament and later ADOS scores in IL-ASD infants (Gomez & Baird, 2005; Zwaigenbaum et al., 2005; Bryson et al., 2007; Clifford et al., 2007; Watson et al., 2007; Garon et al., 2009, 2016). Previous research exploring ER and later ASD symptoms in infant sibling studies have utilized parent-reported measures of early temperament, including the Infant Behavior Questionnaire and Toddler Behavior Assessment Questionnaire, whereas the present study used observational measures of ER, namely heart rate and behavioral affect, suggesting concordance between direct observation and parent report. Furthermore, in line with the present study, Sacrey et al. (2022) utilized observational measures of ER and demonstrated a similar association between early ER and later ADOS-2-T scores, where behavioral affect was predictive of ASD symptom severity in IL infants.

Measures of ER were directly related to later joint attention skills across all participants during the negative and positive phases, with the relation being most pronounced during the negative phases. Specifically, increases in heart rate and negative affect were associated with lower levels of RJA and IJA. Furthermore, the relation was moderated by group, with the IL-ASD group driving the effect. Previous literature exploring the relation between ER and social communication in typically developing children have been mixed. For example, our findings are consistent with Zucal (2010), who examined the relation between parent-reported ER at 15-months and social competence during play with peers at 22 months in typically developing infants. Zucal (2010) found that children who displayed more optimum ER also showed higher social competence when interacting with same-aged peers. However, our findings contrast with those of Cassel et al. (2007), who did not find an association between displays of behavioral affect and joint attention. Cassel et al. (200) investigated the relationship between early

emotional expressions during the Face-to-Face Still-Face Paradigm, a measure of socioemotional regulation, and later RJA and IJA during the ESCS in typically developing children. The discordance between this study and our findings may be due to four important methodological differences. First, ER was assessed at an older age in the present study (12 months in our study vs. 6 months in Cassel et al. (2007) study) and, as such, our participants may have developed more effective regulatory strategies in response to emotionally salient stimuli. Second, we employed both behavioral and physiological indices of ER across several different emotionally salient tasks compared to behavioral measures of affect during one task in the Cassel et al. (2007) study, which may have allowed us to capture more nuanced changes in ER. Third, the ER task in the Cassel et al. (2007) study involved primary caregivers administering the task, whereas the ER task in the present study was completed by an unfamiliar assessor. Finally, the sample in the Cassel et al. (2007) study included only typically developing infants compared to IL and LL children in the present study. Given that the association between ER and joint attention was moderated by the IL-ASD group in our study, the different sample compositions likely contributed to differences in findings. In line with the IL-ASD group in the present study, Nowell et al. (2020) examined the concurrent and predictive relations between regulation (in response to novel sensory stimuli) and joint attention between 13 and 22 months of age, and social communication outcomes at preschool age in children at an increased likelihood for ASD. Although concurrent and predictive relations were found between sensory-regulation and joint attention between 13 and 22 months of age, social communication at preschool age was best predicted by sensory-regulation at 22 months of age. These results highlight that early sensoryregulatory behavior is foundational and influence subsequent social communication development, especially among children at increased likelihood for ASD. Taken together, the findings from these studies emphasize the complexity of the relation between ER and joint attention and how it may be influenced by emerging ASD.

With respect to the relation between joint attention and ASD symptoms, RJA and IJA were inversely related to ADOS-2-T CSS, with low levels of RJA and IJA predicting greater ASD severity. These findings are in accordance with retrospective reports of children later diagnosed with ASD showing abnormalities in joint attention and social orienting behavior by their first birthday (Osterling & Dawson, 1994; Werner, Dawson, Osterling, & Dinno, 2000; Osterling, Dawson, & Munson, 2002), as well as prospective studies of infant siblings (Goldberg

et al., 2005; Yirmiya et al., 2006). For example, as documented in preschoolers with ASD, impairments in RJA have reliably distinguished children with ASD from neurotypical peers and those with developmental disorders (Loveland & Landry, 1986; Baron-Cohen, Baldwin & Crowson, 1997; Sigman et al., 1999; Dawson et al., 2002; Dawson et al., 2004). Mundy er al. (1994) examined joint attention in preschool children with ASD, compared to typically developing children and children with developmental disorders matched by mental age. Consistent with the present findings, joint attention skills were reduced in children with ASD compared to mental age peers. Although the children with ASD in the aforementioned studies were older than the children in our study, the overall pattern and trajectory of joint attention behavior is consistent. The current results, then, add to the growing literature that RJA and IJA in the first two years of life are associated with later ASD symptomatology.

Previous research in neurotypical populations have shown associations between maladaptive ER and subsequent negative outcomes (e.g., poor social relationship, reduced positive affect, increased negative affect, altered physiological arousal; Gross, 2002; Knott et al., 2006; Trepagnier, 1996). Although many of these negative outcomes overlap with ASD symptoms, few studies have investigated the early underpinnings of these associations in infant siblings at IL for ASD. To our knowledge, ours is the first to identify an indirect, longitudinal relations between ER, joint attention, and ASD symptoms, with 12-month behavioral affect related to 18-month RJA, which is related to 24-month ADOS-2-T CSS. These results suggest that RJA mediates the relation between ER and subsequent ASD symptoms for both the negative and positive phases of the ER task. Research on children with ASD has suggested that these processes are linked in early development, where emotional dysregulation is thought to give rise to later ASD symptoms, including impairments in social communication (Rothbart et al., 1992; Garon et al., 2009). Indeed, prospective studies of IL infants who go on to receive an ASD diagnosis, reveal differences in the infant's ability to (1) regulate their emotional states, and (2) initiate joint attention by 12 months of age (Garon et al., 2009; Zwaigenbaum et al., 2013). However, the relation between these processes remains poorly understood. In typical development, ER influences the emergence of social communication. Starting at 4-6 months of age, infants begin to demonstrate voluntary control of emotion, where they start to initiate joint attention with their caregiver to generate a positive state or minimize a negative state (Kopp, 1989). Positive emotionality has been shown to be associated with increases in IJA and RJA,

whereas negative emotionality is associated with reduced IJA (Vaughan et al., 2003; Salley et al., 2007). Thus, early ER appears to play a critical role in the development of social communication and, consequently, may impact the onset of ASD and emerging symptomatology. As such, the relations between affect, RJA, and ASD symptoms may be nuanced and suggests that a complex interplay of ER and joint attention likely contributes to the manifestation of ASD. Given our limited sample, replication of our findings with a larger sample who undergo clinical best estimate diagnoses for ASD is warranted.

Our study has several strengths. We used a multimodal and observational approach to study early ER to examine the developmental relationship between ER and joint attention prior to the age of two. There are some limitations that must be acknowledged, however. First, the results are derived from a sample of siblings of children with ASD and, therefore, may not represent the general population of children with ASD (i.e., non-familial ASD). Second, movement artifacts in the physiological data posed a challenge. Participants who displayed extreme reactions to emotionally valenced stimuli often had associated movement artifacts in the heart rate data, resulting in quality control failure and subsequent data exclusion. Thus, the heart rate data may only include children who have mild-to-moderate reactions to emotionally valenced stimuli. Third, as our study investigated the developmental relations between ER, joint attention, and ASD symptoms, missing physiological and/or behavioral data at the 12, 18, and 24 month timepoints may have limited statistical power to detect differences between IL and LL infants. To this point, due to COVID restrictions, a subset of children were unable to complete the in-person 24-month ADOS-2-T assessment, thus resulting in the use of their 18-month ADOS-2-T scores. As such, replication of these findings in larger samples is warranted. Finally, our modest sample size did not allow for structural equation modeling or cross panel modeling to investigate potential mechanisms between ER, joint attention, and ASD severity. Future studies would benefit from a longitudinal study design with a larger sample of LL and IL siblings.

#### CONCLUSION

In the present study, the examination of the developmental relation between ER and joint attention during the first two years of life highlights the importance and impact of these factors on the emergence of ASD. Our findings indicate that RJA indirectly mediates the relation between ER and later ASD symptoms in children who are at IL for ASD. That is, a complex

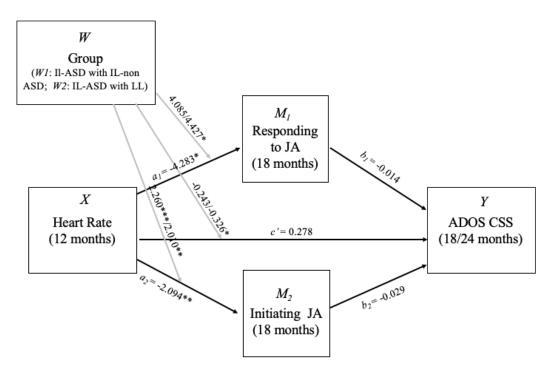
interplay between ER - at behavioral and physiological levels – and joint attention may underlie and contribute to ASD emergence. These findings not only help clarify the nature of ER and joint attention impairments in ASD, but may also inform potential targets and approaches for early intervention for both IL and LL infants who show early signs of ASD.

Table 4.1. Participant	t Demographics
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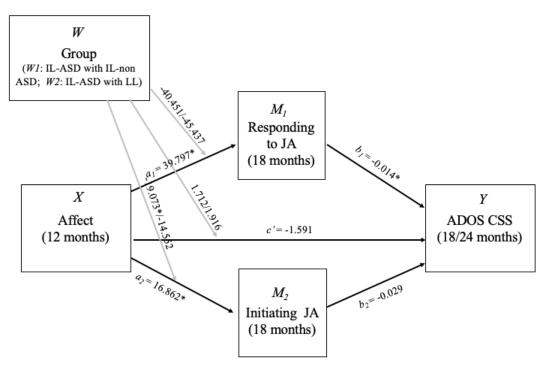
Variable	IL-ASD Infants Mean (SD)	IL-Non ASD Infants Mean (SD)	LL Infants Mean (SD)	Statistic	
n	27	90	54		
Sex (boys:girls)	21:6	42:48	35:19	$X^2 = 10.034$ p = 0.007**	
Negative Phases of ER Paradigm (12 months)					
Mean Affect	-0.420 (0.526)	-0.140 (0.290)	-0.170 (0.361)	H = 5.113 p = 0.078	
Mean Heart Rate	139.650 (8.948)	130.690 (11.000)	130.070 (8.266)	H = 8.691 p = 0.013*	
	Positive Phases of ER Paradigm (12 months)				
Mean Affect	0.030 (0.237)	0.120 (0.312)	0.040 (0.387)	H = 3.140 p = 0.208	
Mean Heart Rate	127.680 (8.791)	125.210 (10.316)	127.060 (9.118)	H = 1.717 p = 0.424	
mESCS (18 months)					
RJA	53.800 (30.088)	79.810 (24.710)	86.300 (19.554)	H = 11.368 p = 0.003 **	
IJA	16.180 (11.007)	23.680 (9.815)	22.060 (9.179)	H= 5.572 p= 0.062	
24 months					
ADOS-2-TCSS	6.930 (2.368)	2.530 (1.486)	2.070 (1.272)	H = 56.626 p < 0.001 **	

*IL-ASD*, infants at increased likelihood with an ASD diagnosis; *IL-non ASD*, infants at increased likelihood but no ASD diagnosis; *LL*, infants at low likelihood of an ASD diagnosis; *mESCS*, modified version of the Early Social Communication Scales; *RJA*, responding joint attention; *IJA*, initiating joint attention; *ADOS-2-TCSS*, Autism Diagnostic Observation Schedule  $-2^{nd}$  Edition – Toddler module Calibrated Severity Score; *SD*, standard deviation.

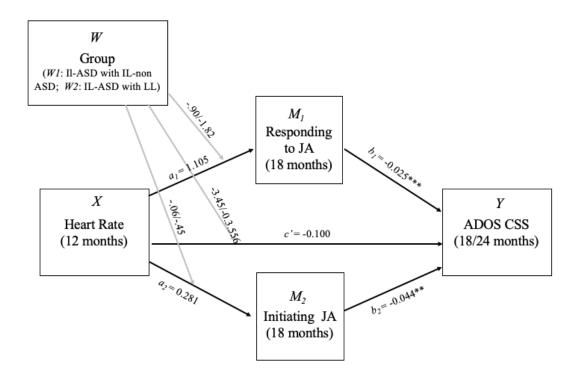
\*p < 0.05, \*\*p < 0.01



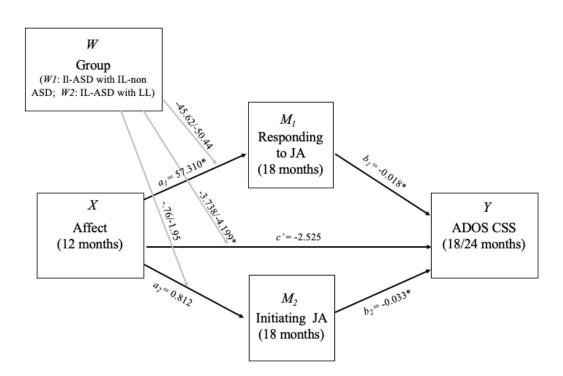
**Figure 4.3.** Parallel mediation with moderation during the negative phases. Model describes RJA  $(M_1)$  and IJA  $(M_2)$  as mediators, Group (W) as the moderator, and Sex as the covariate of the relationship between heart rate (X) and ADOS-2-T CSS (Y).



**Figure 4.4**. Parallel mediation with moderation during the negative phases. Model describes RJA  $(M_1)$  and IJA  $(M_2)$  as mediators, Group (W) as the moderator, and Sex as the covariate of the relationship between behavioral affect (X) and ADOS-2-T CSS (Y).



**Figure 4.5.** Parallel mediation with moderation during the positive phases. Model describes RJA  $(M_1)$  and IJA  $(M_2)$  as mediators, Group (W) as the moderator, and Sex as the covariate of the relationship between heart rate (X) and ADOS-2-T CSS (Y).



**Figure 4.6.** Parallel mediation with moderation during the positive phases. Model describes RJA  $(M_1)$  and IJA  $(M_2)$  as mediators, Group (W) as the moderator, and Sex as the covariate of the relationship between behavioral affect (X) and ADOS-2-T CSS (Y).

### **CHAPTER 5: Discussion & Implications**

The overarching objective of this dissertation was to examine the developmental relationships between early ER and joint attention in infants who are at an increased likelihood (IL) and low likelihood (LL) of an ASD diagnosis. This objective was addressed through three interrelated studies, which addressed the following research questions. First, what are the relationships between early ER and social communication development in childhood? Second, what is the relationship between behavioral affect and physiological arousal during an ER task in a combined cohort of infants who are at an IL and LL of ASD? Third, do variations in behavioral and physiological indices of ER in infancy predict subsequent joint attention in IL and LL infants? The main findings were: (1) as indicated by the systematic review, there was an agerelated bidirectional relationship between ER and social communication in TD children, where associations emerged after six months of age. That is, early ER is critical to the development of social communication, and vice versa; (2) there were no differences in behavior-physiologic congruence between IL and LL infants, suggesting that both groups show similar levels of congruence in response to emotion-eliciting stimuli; (3) there was a relationship between ER and joint attention, for IL infants who were later classified as ASD; (4) there was an indirect relationship between ER and later ASD symptoms, mediated by joint attention, and (5) joint attention was directly related to ASD symptoms across all participants, suggesting that decreased levels of joint attention were associated with increased levels of ASD symptoms. Overall, the findings from this dissertation suggest that there is a developmental relationship between ER and joint attention in infancy, which in turn influences the emergence of ASD.

### 5.1. How Findings Map onto Each Theoretical Framework

The three theoretical frameworks that informed this dissertation include the *Domain-Relevant*, *Temperament*, and *Parallel and Distributed Processing Model* (PDPM). As Chapter 4 explored the relations between ER, joint attention, and ASD symptoms, it will be the main focus of the discussion.

# 5.1.1. Domain-Relevant Framework

The Domain-Relevant framework suggests that functional specialization of emotional and socio-communicative systems is a product of domain-specific (innate) biases, neuronal competition, and environmental experience (Karmiloff-Smith, 2009). A domain-specific framework would suggest that emotional and socio-communicative systems are explained in terms of built-in or innately-specified modules that develop independently of each other, and an interaction between these early-developing systems does not exist. The early capacity for learning, experience, and plasticity cannot be accounted for. The *domain-specific* framework crucially ignores the developmental history of the child and is contrary to the fundamental research question posed by the present dissertation work. In contrast, a domain-general framework posits that experience imprints itself via general learning mechanisms, where plasticity is the sole mechanism for the development of neural systems and networks. Adopting this framework in its extreme suggests that emotional and socio-communicative systems may develop non-concurrently (i.e., it is dependent on environmental experience) and in a lessstructured manner. Consequently, this may suggest less inherent relationships between ER and joint attention and that early competencies or early deficits identified in IL infants is not easily explained by the *domain-general* framework, given that this position argues that experience affects all systems across the board in the same manner (rather than at a distinct level or systemlevel). The Domain-Relevant framework, however, provides a more conceptually coherent and empirically sound foundation than the *domain-specific* or *domain-general* frameworks. The findings from this dissertation supports the Domain-Relevant framework.

Support for the *Domain-Relevant* framework comes from the systematic review of the relationships between ER and social communication (Chapter 2). Review findings suggest that ER and social communication are reciprocal, where young children's regulation of emotions is vital to the development of social communication and vice versa. This pattern appears to become more pronounced with age, but there is tremendous heterogeneity in how these constructs were defined and measured in the literature. Moreover, the relationship between ER and social communication is likely more complex and nuanced rather than a direct one-to-one relationship. The results from Chapter 4 also lend support for the *Domain-Relevant* framework, where ER and joint attention were significantly related to each other. Findings suggested that emotional dysregulation was associated with lower levels of IJA and RJA (with the caveat that this relationship was only examined in one direction; ER to IJA/RJA). Together, the pattern of

findings suggest that ER and joint attention have a reciprocal relationship, with each directly or indirectly impacting the other.

### 5.1.2. Temperament Framework

The *Temperament* framework can be viewed as a specific model of the *Domain-Relevant* framework. While the *Domain-Relevant* framework is applicable to two or more developmental processes, the *Temperament* framework narrows in on two interrelated processes. That is, joint attention may be influenced by emotional-processing systems (i.e., defense or approach systems), including the child's reactive tendencies to express and experience emotions (Rothbart, 2011), and may have implications for subsequent social-communicative behavioral responses. The converse is also plausible, where early control of orienting attention (i.e., early form of joint attention) influences ER, thereby guiding social interactions. As an infant matures and develops, he/she gains considerable control over this process and associated ER. That is, orienting of attention is refined and becomes a voluntary, independently implemented self-regulatory strategy with a social partner. This may have implications for future social interactions. Overall, within the *Temperament* framework, one would expect that emotional and socio-communicative processes are mutually dependent on each other and work together to stabilize internal and external experiences. Consequently, this would shape the infant's internal representation and external interaction with the social world.

Support for the *Temperament* framework comes from the systematic review (Chapter 2), in which the majority of the ER constructs were derived from questionnaires that measured temperament. For example, 6-month negative affect (as measured by the Infant Behavior Questionnaire – Revised) was associated with 12-month coordinated attention (Markova, 2008) and 12-month activity level, distress to limitations, and fear (as measured by the Infant Behavior Questionnaire) were associated with 19-month sociability (as measured by a stranger sociability procedure; Thompson & Lamb, 1982). More broadly, the findings from Chapter 2 support the perspective that temperament contributes to the formation of a range of socio-communicative abilities, with the strength of the relationship increasing with age. In concordance, the findings from Chapter 4, which employed sub-tasks from the Laboratory Temperament Assessment Battery (Goldsmith & Rothbart, 1996), revealed a direct relationship between observed measures of ER (affect and heart rate) and later joint attention skills. Specifically, both behavioral and physiological indices of ER were directly related to subsequent IJA and RJA during the

negatively valenced tasks. That is, increased heart rate and increased displays of negative affect at 12 months of age were associated with decreased IJA and RJA at 18 months, suggesting that emotional dysregulation impacts later social communication development regardless of sibling status. The findings from these dissertation studies lend stronger support for the *Temperament* framework, which focuses on the directional relationship that ER plays in the emergence of social communication skills versus the *Domain-Relevant* framework, which posits a broader bidirectional relationship. That is, this dissertation did not adequately examine the converse relationship (i.e., social communication influencing later ER).

# 5.1.3. Parallel and Distributed Processing Model (PDPM) Framework

The *PDPM* framework suggests a parallel, reciprocal relationship between ER and joint attention (Mundy et al., 2009, 2010). That is, the development and maturation of joint attention and emotion systems interact in a reciprocal cause-and-effect manner with experience, which may, in turn, have cascading effects on the development of later skills. The *PDPM* framework provides two important insights. First, although the *PDPM* framework focuses on the relationship between joint attention and later social competence, early ER also contributes to the development of both processes. Second, the *PDPM* framework predicts RJA and IJA may be differentially influenced by the same factor.

The empirical study presented in Chapter 4 provides support for the *PDPM* framework, as an indirect, longitudinal relationship between ER, joint attention, and ASD symptoms emerged in IL and LL infants. Early ER was differentially related to RJA, but not IJA, with RJA further related to later ASD symptoms. In other words, RJA mediated the relationship between ER and ASD symptoms, where emotional dysregulation was associated with lower levels of RJA, which was then associated with higher ASD symptom severity.

### 5.1.4. Summary

Each of the three theoretical frameworks were supported by the results of this dissertation. The findings from each chapter, however, do not lend support to one framework over the others. That is, because the directional relationship between early ER and later joint attention was explored, we are unable to comment on the converse relationship (i.e., early joint attention and subsequent ER). Future work that examines the bidirectional and longitudinal

relationship between ER and joint attention, as well as their relationship with emerging ASD symptoms, is warranted.

### 5.2. Limitations

There are a number of important limitations to the dissertation studies. As with many prospective infant sibling research studies (Zwaigenbaum et al., 2005), the primary limitation was small sample size. As COVID-19 restrictions were put into place during the midst of our data collection, a large subset of children were unable to complete in-person ER, joint attention, and ASD assessments. This proved difficult when investigating the bidirectional relationship between ER and joint attention, as we were only able to examine the ER to joint attention directional relationship (not the joint attention to ER relationship) due to incomplete data. Furthermore, not all infants were able to complete their 36-month ASD diagnostic assessments, the age at which a diagnosis is more stable. As we used the 24-month ADOS-2-T to characterize ASD symptoms, it is unknown whether these infants will consistently meet criteria for ASD. As such, our description of early symptoms of ASD is exploratory and requires follow-up.

Additionally, the small sample size led to analytical difficulties. That is, it decreased overall statistical power to detect group differences and longitudinal relationships. The analyses in the dissertation studies utilized correlations and parallel mediation with moderation analysis to examine the relationships between ER, joint attention, and later ASD symptoms. However, multivariate techniques to evaluate multiple relationships between variables (such as structural equation modeling or cross panel modeling) were unable to be conducted due to modest sample size. As a result, the present findings should be interpreted with caution.

There were also methodological limitations. First, novel to this research was physiological data collection during the ER task. As heart rate was measured at 6, 12, and 18 months of age, it is possible that movement artifacts during the ER task reduced the quality of physiological data. For example, infants with extreme reactions to emotion-eliciting stimuli and/or were mobile during the task often had movement artifacts displayed in their heart rate data. This led to failure in quality control, resulting in "messy data" and exclusion of these data. Exclusion of these data not only reduced the sample size (i.e., making analysis with matched data difficult) but also may have yielded a sample of IL and LL participants with muted or mild responses during the ER task. As such, findings may be based on smaller sample sizes with less variability in physiology.

### **5.3.** Clinical Implications and Future Directions

Identifying early risk markers of ASD in infancy can inform the developmental course of symptom emergence and severity over time. Rogers (2009) indicated that many early case studies of ASD reported secondary symptoms - such as irritability, poor motor skills, and sensory responsivity - appearing before core symptoms of ASD. This suggests that ASD not only affects the core diagnostic domains (as outlined in the DSM-5), but multiple domains of development. In fact, prospective studies of IL infants reveal differences in the infant's ability to regulate emotional states and initiate joint attention as early as 12 months of age (Garon et al., 2009; Zwaigenbaum et al., 2013). Therefore, examining early markers such as ER and joint attention that may moderate or overlap with ASD symptoms is crucial to better understand the underlying mechanisms and pathways leading to ASD emergence.

The present dissertation research examined early symptom development by focusing on the relationship between ER and joint attention as early markers of ASD. By utilizing behavioral and physiological data - not previously reported (Adrian et al., 2011) - the developmental pathways by which ER and joint attention emerges in IL infants were examined, as well as how these processes give rise to later ASD symptoms. While our behavioral and physiological measures of ER may not be feasible for use in clinical practice at the present time, it will be important to continue to expand this work to determine if these measures are indeed reliable markers of early ER in IL and LL infants. Furthermore, additional research examining the longitudinal, bidirectional relationships between ER and joint attention may further illuminate ASD symptom presentation and emergence. That is, investigating these relationships can improve our understanding of the mechanisms underlying ASD etiology, phenotypic overlap between core diagnostic symptoms, and comorbid psychopathologies, as well as clarify the role ER and joint attention play in functional outcomes of individuals with ASD. This may contribute to early identification of infants likely to develop ASD, but also inform the development of interventions that can be applied clinically. For example, by focusing on early aspects of ER and joint attention, the 'Social ABCs' intervention model (Brian et al., 2017) - an efficacious, parentmediated intervention in 16- to 30-month old children with early signs of ASD – can be adapted

for younger at-risk infants. Thus, this research has the potential to not only shed light on mechanisms underlying early deficits in ASD, but has broader implications for informing treatment targets to improve outcomes in affected children.

Future research is needed to replicate these findings and should be completed with a larger sample. That is, adequately powered longitudinal studies are needed to elucidate how atypicalities in ER and joint attention may increase the risk for ASD and other comorbid psychiatric disorders. In fact, growing evidence suggests that impairments in socio-emotional development may limit functional outcomes for children with ASD and give rise to a range of co-occurring and/or comorbid problem behavior (e.g., anxiety, ADHD; Gadow et al., 2004; Brereton et al., 2006; Witwer & Lecavalier, 2008). Up to 50% of children with ASD meet clinical criteria for two or more mental health disorders, the underpinnings of which appear in early childhood (Raza et al., 2019; Kiss et al., 2017). As such, studies following both IL and LL infants and early diagnosed children into adolescence and beyond may provide a clearer picture of trajectories, as well as identify clusters of behavior that are predictive of comorbid diagnoses. Finally, continuing to integrate information across several methodologies and assessments (i.e., combinations of behavioral observation, physiological measurement, and parent-report) may provide a more holistic picture of the ER and joint attention processes.

# 5.4. Conclusion

In summary, the present work examined the developmental relationship between ER and joint attention in a sample of infants at an IL and LL for ASD. Furthermore, this relationship was negatively influenced by the emergence of ASD, suggesting a complex interplay between these processes in young children who are at IL for ASD. These findings are consistent with the view that early skills, such as ER and joint attention, have cascading and additive effects that may influence later developmental outcomes. As such, early detection and improved understanding of early socio-emotional atypicalities, especially in young children at IL for ASD, is warranted to promote healthy outcomes and competencies.

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# APPENDIX A

## Search Strategy for Chapter 2 (Systematic Review)

Ovid MEDLINE(R) and Epub Ahead of Print, In-Process & Other Non-Indexed Citations and Daily 1946 to March 20, 2022 (OVID Interface)

- 1. joint-attention.mp.
- 2. communication/ or nonverbal communication/
- 3. social behavior/ or social skills/
- 4. social communication.mp.
- 5. (social\* or communicat\*).ti.
- 6. (social\* or communicat\*).ab. /freq=2
- 7. 1 or 2 or 3 or 4 or 5 or 6
- (emotion\* regulation or emotion\* management or emotion\* competence or affect\* regulation).mp.
- 9. (emotion\* or affect or affective or temperament\*).mp.
- 10. 8 or 9
- 11. 7 and 10
- 12. limit 11 to "all infant (birth to 23 months)"
- 13. 11 and (infan\* or neonat\* or toddler\* or newborn\*).mp.
- 14. 12 or 13
- 15. limit 14 to animals
- 16. 14 not 15
- 17. (feeding or breastfeeding or pregnan\* or postpartum or post-partum or post-natal or postnatal or nutrition or social media or monk\* or pig\*).ti.
- 18. 16 not 17
- 19. Epidemiologic studies/ or exp case control studies/ or exp cohort studies/ or Crosssectional studies/
- 20. (Case control or cohort or Follow-up or (observational adj (study or studies)) or Longitudinal\* or Retrospective\* or Cross sectional\*).tw.
- 21. 18 and (19 or 20)

22. (trajector\* or "each age" or "across infanc\*" or "across age\*" or "across time" or "over time" or "time points" or "developmental course" or age-related or "remain\* stable").ab.
23. 21 or (22 and 18)

### PsycINFO 1806 to February Week 4 2022 (OVID Interface)

- 1. joint-attention.mp.
- 2. communication/ or exp nonverbal communication/ or communication skills/
- 3. social behavior/ or social skills/
- 4. social communication.mp.
- 5. (social\* or communicat\*).ti.
- 6. (social\* or communicat\*).ab. /freq=2
- 7. 1 or 2 or 3 or 4 or 5 or 6
- (emotion\* regulation or emotion\* management or emotion\* competence or affect\* regulation).mp.
- 9. (emotion\* or affect or affective or temperament\*).mp.
- 10. 8 or 9
- 11. 7 and 10
- 12. 11 and (infan\* or neonat\* or toddler\* or newborn\*).mp.
- 13. limit 12 to animal
- 14. 12 not 13
- 15. (feeding or breastfeeding or pregnan\* or postpartum or post-partum or post-natal or postnatal or nutrition or social media or monk\* or pig\*).ti.
- 16. 14 not 15
- 17. (Case-control or cohort or Follow-up or (observational adj (study or studies)) or Longitudinal\* or Retrospective\* or Cross sectional\* or prospective\*).tw.
- 18. (trajector\* or "each age" or "across infanc\*" or "across age\*" or "across time" or "over time" or "time points" or "developmental course" or age-related or "remain\* stable").ab.
- 19. 16 and (17 or 18)

# SCOPUS (Scopus Interface)

(TITLE-ABS-KEY(joint-attention OR nonverbal-communication OR communication-skills OR social-behavior OR social-skills OR social-communication) OR TITLE(social\* OR communicat\*)) AND TITLE-ABS-KEY(emotion\* or affect or affective or temperament\*) AND TITLE-ABS-KEY(infan\* or neonat\* or toddler\* or newborn\*) AND (TITLE-ABS-KEY(infan\* or neonat\* or toddler\* or newborn\*) AND (TITLE-ABS-KEY(epidemiolog\* or case-control or cohort or follow-up or observational\* or longitudinal\* or retrospective\* or cross-sectional\* or prospective\*) OR ABS (trajector\* or "each age" or "across infanc\*" or "across age"\* or "across time" or "over time" or "time points" or "developmental course" or age-related or "remain\* stable")) AND NOT TITLE(feeding or breastfeeding or pregnan\* or postpartum or post-natal or postnatal or nutrition or social-media or monk\* or pig\*)

# CINAHL Plus with Full Text (EBSCO Interface)

((MH "Communication") OR (MH "Communication Skills") OR (MH "Nonverbal Communication+") OR joint-attention or nonverbal-communication or social-communication or social-behavior or social-skills OR TI (social\* or communicat\* )) AND (emotion\* or affect or affective or temperament\* ) AND (infan\* or neonat\* or toddler\* or newborn\* ) NOT TI ( feeding or breastfeeding or pregnan\* or postpartum or post-partum or post-natal or postnatal or nutrition or social-media or monk\* or pig\* ) AND ( (MH "Nonexperimental Studies+") OR Epidemiologic\* or longitudina1\* or case-control or cohort or cross-sectional\* or follow-up or observational or retrospective\* or prospective\* OR AB ( trajector\* or "each age" or "across infanc\*" or "across age\*" or "across time" or "over time" or "time points" or "developmental course" or age-related or "remain\* stable" ))

# **APPENDIX B**

Brief Coding Scheme for Events and Affect for the Emotion Regulation Task

Events					
(continuous, mutually exclusive & exhaustive)					
Code		Description			
Baseline Video 1, 2, or 3	From start to end of baseline video: code as number 1, 2 or 3 as				
	they occur	they occur			
Bubbles	Experimenter (E) is blowing bubbles				
Toy Play	E places toy on table	directly in front of infant and allows infant			
	to play	to play			
Toy Removal	E lifts toy to hold out of reach but in view of infant.				
Blank mask	E is wearing blank mask				
Cow mask	E is wearing cow mask				
Wash Face	E washes infant face with wet wipe				
Brush Hair	When E is brush infa				
Event Transition/other	All events outside those described above.				
Affect					
(instantan	eous in 5 s intervals, mutua	lly exclusive and exhaustive)			
Codes	Modifiers	Description			
Affect not codable		No cues to affect for the entire 5 s			
Affect not codable	-	interval			
Transition	-	5s interval does not contain coded events			
Affect Codable	2	Extreme positive affect			
	1	Mild to moderate positive affect			
	0	Neutral affect			
	-1	Mild to moderate negative affect			
	-2	Extreme negative affect			