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Role of the Vagal System in the Regulatory Sensory Processing Patterns in Children with Autistic Spectrum Disorder: Physiologic Underpinnings and Reliability of Measurement

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ROLE OF THE VAGAL SYSTEM IN THE REGULATORY SENSORY PROCESSING PATTERNS IN CHILDREN WITH AUTISTIC SPECTRUM DISORDER: PHYSIOLOGIC UNDERPINNINGS AND RELIABILITY OF MEASUREMENT

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Abstract

ROLE OF THE VAGAL SYSTEM IN THE REGULATORY SENSORY PROCESSING PATTERNS IN CHILDREN WITH AUTISTIC SPECTRUM DISORDER: PHYSIOLOGIC UNDERPINNINGS AND RELIABILITY OF MEASUREMENT

by

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This study was a preliminary investigation into the reliability of cardiac vagal tone (CVT) as a measure of regulatory sensory processing in typically developing children and children with autistic spectrum disorders (ASD).

We also examined group differences in CVT at rest and in response to sensation and the relationship of CVT to behavioral measures of sensory processing. The goals of this study were to answer three related research questions: 1. Is CVT a reliable measure of regulatory sensory processing? 2. Do children with ASD respond to sensory information differently than typically developing children? 3. Is there a relationship between physiologic and behavioral measures of regulatory sensory processing?

The participants were 15 typically developing boys and 11 boys with ASD. We measured CVT two times, within 4 weeks, during the Sensory Challenge Protocol (Miller, 1999). Behavioral responsivity to sensation was measured using the Short Sensory Profile (SSP) (Dunn, 1997). Reliability was assessed using intraclass correlation coefficients, vagal response patterns were assessed using repeated measures analysis of variance and t-tests and multivariate analysis of variance were used to look at between group differences in vagal measures and SSP scores.

Pearson Correlation coefficients were used to examine relationships between CVT and SSP scores.

Results of this study indicate that CVT is a reliable measure of regulatory sensory processing. Highlighting the fact that the children with ASD demonstrate greater statistical reliability than the typically developing children. We also found that the children with ASD demonstrate significantly lower baseline vagal tone. Further, we found the two groups responded differently to sensation. Post hoc tests revealed a significant difference between the groups on vagal responsivity to vestibular stimulation, with the group of children with ASD demonstrating less reactivity. Additionally, the ASD group demonstrated significantly lower scores on all sections of the SSP except movement sensitivity. Lastly, several correlations between vagal responses during the SCP and SSP sections were noted.

The results of this study point to a relationship between physiology and behavior, suggesting that children with ASD demonstrate less physiologic flexibility which may play a role in the decreased behavioral flexibility seen in this population.

Chapter I

Introduction

Background

The Diagnostic and Statistical Manual, Fourth edition, characterizes autism spectrum disorders (ASD) as having marked impairments in three categories: social interaction, communication and stereotyped patterns of behaviors. Of the three, the least is known about the stereotyped patterns of behaviors which in include "inflexible adherence to non-functional routines, pre-occupation with at least one restricted pattern of interest, and stereotyped or repetitive motor mannerisms" (American Psychiatric Association, 1994). While little is known about the reasons for these rigid inflexible patterns of behavior characteristic of children with ASD, there is speculation that they may be sensory driven.

The literature on behavioral responsivity to sensation in children with autism indicates that their behavioral response patterns are significantly different than children without autism (Kientz & Dunn, 1996; Watling, Dietz, & White, 2001). It is estimated that 70-90% of individuals with autism show atypical patterns of sensory processing that interfere with occupational performance (Adrien, Ornitz, Barthelemy, Sauvage, & Lelord, 1987; Eaves, Ho, & Eaves, 1994; Kientz & Dunn, 1996). Baraneck (1999) found that children with ASD showed early signs of deficits in sensory motor processing as evidenced by first year video tape analysis suggesting that sensory processing problems may be a core feature in children with ASD.

Sensory modulation is defined as the capacity to regulate and organize the degree, intensity and nature of responses to sensory input in a graded and adaptive manner (Miller & Lane, 2000). This allows an individual to maintain an optimal range of performance and adapt to challenges in daily life. Efficient modulation of sensory input is essential for self-regulation and adaptive interactions with our environment. "Sensory input can facilitate or impede self-regulating responses and behavior" (Reeves, 2001). Sensory Modulation Disruption (SMD) is an inability to regulate the manner in which our brains and bodies process and respond to sensory information and results in a difficulty adapting to the challenges of daily life and occupational performance. SMD includes hyper-responsivity, hypo-responsivity and fluctuating responsivity (Miller & Lane, 2000). SMD may occur as a primary disorder or in conjunction with other disorders such as autism spectrum disorders. While there is a good deal of literature on the behavioral response patterns to sensation of children with ASD very little is known of the physiologic response patterns to sensation. Currently, there is a group of researchers from universities around the country who are investigating the physiologic responsivity of various groups of children with proposed disruptions in sensory processing. They call themselves the Sensory Integration Research Collaborative (SIRC) and have provided some preliminary information on how children with autism physiologically respond to sensation. The physiologic measure used thus far in the studies involving children with ASD is electrodermal reactivity (EDR), which will be explained later in this paper. They found that children with autism demonstrated EDR patterns that were different than typically developing children and different than children in other diagnostic groups such as attention deficit disorder (McIntosh, Miller, Shyu, & Hagerman, 1999; Donelan-Mangeot et al., 2001; Miller, McIntosh, Reisman, & Simon, 2001; Shoen, S.A., Miller, L.J., Brett-Green, B., & Hepburn, S.I., 2008). Specifically, they found that children with ASD demonstrate decreased magnitude EDR's in response to sensation indicating a hypo-responsivity to sensation (Miller, McIntosh, Reisman, & Simon, 2001). When looking specifically at children with high functioning autism and asperger's syndrome it was found that this group could be divided into two subgroups based on the EDR patterns in response to sensation, a high arousal group as evidences by EDR's of greater magnitude and a low arousal group as evidenced by depressed magnitude of EDR. (Shoen et al., 2008).

Theoretical Perspective

Neuro-occupation is a theoretical perspective that seeks to explain the relationship between neural processes and human behavior (Padilla & Peyton, 1997). As individuals engage in the activities to fulfill their daily roles and responsibilities (occupations) the interactions and events encountered result in neural adaptations that affect subsequent behavior. Thus, human behavior influences, and is influenced by, the dynamic interaction between daily events and neural processes. The autonomic nervous system (ANS) is thought to provide the foundation for our behavior in that it regulates physiologic responses to both internal and environmental demands which supports our ability to engage in activities and interactions (Koizumi, 1991) and is thus foundational to occupational performance.

The ANS regulates the neurophysiologic changes necessary to meet both internal (body) and external (environmental) demands. These neurophysiologic changes enable us to physically and mentally respond to a situation and then return our bodies to a resting state. However, it is not only the transient shifts in internal activity that allow for adaptive functioning but also level of resting , or basal, physiologic state of an individual as this is the baseline from which theses neurophysiologic shifts occur . The flexibility of this basal physiologic state can influence the effectiveness of the shifts needed for adaptive functioning within an ever-changing environment (Porges, 1992). Individuals with less physiologic flexibility are thought to have less behavioral flexibility when responding to environmental demands (Porgcs, 1995). A characteristic trait of individuals with autism spectrum disorders is behavioral rigidity in responding to environmental demands. This inflexibility may be due to the need to maintain the "safety" of one's environment by controlling the types of sensation encountered in that environment. Individuals with autistic spectrum disorders frequently show atypical behavioral responses to sensation which may interfere with the ability to respond to the environment in an adaptive manner. Based on the theory of neuro-occupation the question arises as to what neuro-physiologic processes may be underlying the behavioral inflexibility in responding to sensation seen in children with ASD?

Regulatory Disorders

In addition to disruptions in sensory processing evidence suggests that children with autism have "regulatory disorders with diminished ability to modulate arousal." (Heubner, 2001). Self-regulation may be defined as the ability to adjust to changing conditions through internal processes and behaviors to maintain a sense of control (Reeves, 2001). Regulatory disorders are characterized by difficulties in regulating behavior and responses to sensory, motor and affective input in order to organize a calm, alert or positive affective state. Symptoms include difficulty in calming, affective lability, poor attention, difficulty making transitions, impulsivity, hypersensitivities to sensory stimulation and behavioral patterns of avoidance or withdrawal (Heubner, 2001). Self- regulation is foundational to adaptive functioning within one's environment. Bagnato and Neilsworth (1999) found that children with autism, as compared to children with developmental delays and fragile x syndrome, demonstrated the most severe regulatory disorders. In a study by Willemsen-Swinkles, S.H.N., Buitelaar, J.K., Dekkar, M. & vanEngland, H. (1998) a high physiologic arousal state (a component of regulatory disorders) was found to precede the onset of stereotypic sensorimotor behaviors such as hand flapping, in 85% of the instances, which may suggest that behaviors so often seen in children with autism may be related to a physiologic process.

Both empirically and anecdotally, we know that children with autism demonstrate many, and collectively all, of the behaviors associated with disorders of self-regulation and sensory modulation, and that these two disorders have a great deal of overlap. Therefore, in an effort to clarify terminology and increase effectiveness in diagnosing and treating children with both SMD and regulatory disorders a task force was organized to re-classify these disorders. The result of this re-classification was the establishment of unified term for disorders of regulation and modulation which is regulatory sensory processing disorders (RSPD) (Interdisciplinary Council on Developmental and Learning Disorders, 2005).

Regulatory-Sensory Processing

In order to clarify the terminology that will be of use throughout this paper a brief review the new classification system for disorders of regulation and sensory modulation now collectively referred to as Regulatory Sensory Processing Disorders is warranted. In the past, children demonstrating atypical behavioral responses to sensation were diagnosed with one of two disorders, a sensory processing disorder (SPD) or a regulatory disorder. Occupational Therapists for many years have been working under a Sensory Processing framework, originally proposed by Jean Ayres in 1972. The framework suggests that behavior is driven by the underlying neurophysiologic processes involved in sensory processing. Occupational Therapists utilizing a sensory processing framework often labeled children who had difficulties with calming, difficulties with transitions, affective lability and hyper and hypo responsivities to sensation as having a disorder of sensory processing. In contrast, psychologists working with children often follow the work of Stanley Greenspan and his Developmental, Individual-Difference, Relationship- Based (DIR) model of infant and early childhood mental health from which the classification of Regulatory Disorders emerged. The DIR model focuses on a child's social and emotional capacities and relationship patterns and takes into account individual differences in processing information, including sensory processing, that contribute to the emotional and social patterns that a child develops. Psychologists and other professionals working under the DIR model label children with difficulties described above as having a regulatory disorder. Thus, depending on the professional's frame of reference, a child with difficulties with calming, transitioning, affective lability and hyper and hypo responsivity to sensation was labeled as having either sensory processing disorder or regulatory disorder.

The two schools of thought however were not mutually exclusive. Many Occupational Therapists practicing in early intervention also work under a DIR model of intervention for children with regulatory and sensory processing disorders. Similarly, many psychologists believe in the tenets of sensory processing theory and utilize compensatory strategies from this framework in treatment. The overlap between the perspectives made it difficult to clearly distinguish the differences between sensory processing disorders and regulatory disorders. As such, a team of individuals, leaders in the field of regulatory and sensory processing disorders, formed a work group in 2005 to dissect the two disorders. The panel is now known as the Regulatory-Sensory Processing Work Group of the Interdisciplinary Council on Development and Learning Disorders (ICDL). The work group brought these two seemingly separate schools of thought together and conceptually merged their ideas. The unified label for disorders characterized in part by difficulties with calming and transitions, affective lability and hyper and hypo responsivity to sensation is now Regulatory-Sensory Processing Disorder (RSPD).

Regulatory-Sensory Processing (RSP) refers to the way in which sensory information coming from the body and the environment is perceived and organized to produce an adaptive response (Interdisciplinary Council on Developmental and Learning Disorders, 2005). According to this definition adaptive responses are both the behavioral response to sensation as well as the physiologic response to the information. Regulatory-Sensory Processing Disorders (RSPD) are characterized by difficulties with emotions, behaviors and motor abilities in response to sensory stimulation that leads to an impairment in development and function. Regulatory-Sensory Processing Disorders may be diagnosed as a primary disorder or in conjunction with other disorders such as attention deficit hyper-activity disorder and autism. Hence, it is reasonable to speculate that symptoms of RSPD may be at the root of the behavioral and interactive difficulties seen in children with autistic spectrum disorders.

The RSPD workgroup designed a classification system to help families and clinicians better understand the behavioral symptoms seen in various populations of children including children with autism. The new classification of Regulatory –Sensory Processing Disorders in the Diagnostic Manual for Developmental and Learning Disorders defines several sub-groups within this diagnosis that help clinicians and families make sense of certain patterns of behavioral responses which may have otherwise appeared disconnected.

There are three main types of RSPD, Sensory Modulation Challenges (Type I), Sensory Discrimination Problems (Type II) and Sensory-Based Motor Challenges (TypeIII). There is also a mixed type, which includes aspects from all three main types. Under each of these primary types there are several sub-types. Please see Appendix A for classification system.

While the theoretical perspective behind the classification system for RSPD recognizes that there are neurophysiologic underpinnings for these behavioral response patterns it does not, and cannot, identify the specific neurophysiologic profiles due to the scarcity of research in this area. However, the body of research on the physiologic correlates of the behavioral symptoms seen in RSPD is growing fast and researchers are hoping to identify specific patterns of neurophysiologic reactivity which may help us better define and treat the disorder.

Measures of Autonomic Function

Currently, researchers in the Sensory Integration research Collaborative, and others, are using two physiologic indexes to measure responsivity to sensation, electrodermal activity and cardiac vagal tone. Both serve as indirect measures of ANS functioning. Electrodermal activity (EDA) is an indirect measure of sympathetic nervous system function. The main function of the SNS is to prepare and defend the body in response to severe external challenges or stressors. The SNS is referred to as the "fight or flight" system. At rest, and in the presence of external events that are perceived as benign, the SNS is not active or is minimally active. On the other hand, the SNS is fully active during events perceived as stressful by the individual.

Electrodermal activity refers to the changes in the electrical conductance of the skin associated with eccrine sweat gland activity, which are under SNS influence, and has two components, skin conductance level (SCL) and electrodermal responsivity (EDR). Skin conductance level is the tonic level of conductivity of the skin. Electrodermal responsivity is the phasic changes that occur in response to specific stimuli. Electrodermal responses occur in the presence of startling or threatening stimuli. Research involving electrodermal responsivity has been conducted on various populations of children with proposed disruptions in sensory modulation including fragile x, attention deficit disorder, with and without hyperactivity, primary sensory modulation disorder and autism spectrum disorders. In aggregate, the findings tell us that each group demonstrates EDR patterns that are different than typically developing children and that each group demonstrate patterns that are different from each other (McIntosh, Miller, Shyu, & Hagerman, 1999; Miller, McIntosh, McGrath, Shyu, Lampe, & Taylor, 1999; Donelan-Mangeot et al., 2001; Miller, L.J., McIntosh, D.N., Reisman, J.E., Simon, J., 2001). In particular, children with autism spectrum disorders demonstrated hypo-responsivity to sensation as evidenced by a decreased magnitude of EDR's as compared to typically developing children. However, the sample size in this study was small and the authors caution interpretation of results. While the information on SNS activity in response to sensation is important in providing us with preliminary information on the autonomic nervous systems role in sensory processing, it does not provide us with information on the basal supportive function of the nervous system and how that can impact neural activity in response to sensation, which is the function of the PNS. Therefore, the remainder of this paper will focus on the parasympathetic nervous system's role in regulatory -sensory processing.

The second measure being used is cardiac vagal tone (CVT). CVT is the measure of interest in this study and therefore will be focus for the remainder of this paper. Cardiac vagal tone is an indirect measure of parasympathetic nervous system function. The primary role of the PNS is to maintain homeostasis. Homeostasis is a dynamic process in response to both internal needs and perceived or actual external demands. It involves a constant fine-tuning of multiple metabolic and physiologic systems to attend to an external challenge and to return an organism back to a state of stability after disruption. At rest we typically have a high PNS tone which allows the body's restorative functions to take place. When an event occurs, a disruption of this resting state is necessary to meet the demands of the event and to support appropriate engagement and disengagement with the environment (Porges, Doussard-Roosevelt, Portales, & Greenspan, 1996). This responsive state is typically defined by the withdrawal of PNS tone. Thus, regulation of PNS activity is required in each and every situation that a person encounters in daily life. At rest the PNS is highly active creating an environment where the physiologic processes necessary for the body's restorative functions to occur can be achieved. While a PNS withdrawal is necessary when a person needs to engage with the environment. The SNS on the other hand is only called to activation in stressful situations. Therefore investigation into the PNS's role in behavior may provide us with valuable information on the nervous system's supportive role in shaping behavior. For example engaging in a social interaction (not typically a stressful situation) requires certain physiologic changes, such as increased heart rate and decreased digestive functions which require a withdrawal of PNS tone without a rise in SNS activity.

Cardiac vagal tone is a commonly used measure of PNS functioning (Schaaf, Miller, Seawell, & O'Keefe, 2003; Schaaf et al., 2010; Porges,1995; Porges et al., 1996; Suess, Porges and Plude, 1994; Boccia and Roberts, 2000; Calkins, 1997; DiGangi, Greenspan, & Porges, 1991).Cardiac vagal tone is a measure of the nervous system's modulation of heart rate via the vagus nerve. This process reflects a continuous feedback loop in between neural processes and environmental demands. Cardiac vagal tone is indexed in the amplitude of the respiratory sinus arrhythmia. Respiratory sinus arrhythmia is the rhythmic heart rate variability, within a normal range, associated with spontaneous breathing, with heat rate typically increasing with inhalation and decreasing with exhalation (Porges, 1995). RSA occurs from inhibition of PNS control therefore measuring its amplitude is a good indicator of PNS tone, specifically vagal influences over heart rate.

The Vagal System

The vagus is the tenth cranial nerve. However, unlike other cranial nerves, which are thought to be primarily effective in nature, the vagus is both effective and affective. It sends and receives messages to and from the viscera in order to maintain physiologic stability when one is at rest or when faced with an external demand. Implicit in this vagal system are motor pathways to change visceral state, sensory pathways to monitor visceral state and brain structures involved in the evaluation of input and regulation of output (Porges, 2005).

Based on this description, it seems as though the vagal system plays a large role in regulatory sensory processing. However, the specific patterns of vagal responsivity in both typically developing children and children with autism remain unclear.

The Polyvagal Theory as proposed by Porges (1995) describes a model of neural regulation of the ANS that highlights two functions of the vagal system. The first function is to maintain a homeostatic balance of the internal viscera to enhance growth and restoration in the absence of environmental demands (at rest). This is controlled by the unmyelinated branch of the vagus nerve whose fibers originate in the dorsal motor nucleus. The second function is to provide rapid adjustments of neurophysiologic processes by regulating heart rate via the sino-atrial node and is controlled by the myelinated branch of the vagus nerve whose fibers originate in the nucleus ambiguous. This is usually observed as a suppression of vagal tone when one is faced with an event, otherwise known as the *vagal brake* (Porges et al, 1996). The suppression of vagal tone in response to an external demand allows attention and resources to be directed outward, which allows the individual to meet the needs (social, cognitive, motor) of the situation. If the vagal system is not suppressed the physiologic shifts necessary for motor, social and cognitive responses are not initiated. As mentioned above, individuals with less variability are thought to have less physiologic flexibility which may result is less behavioral flexibility (self-regulation). Therefore, the less variability in vagal tone (low vagal tone), the less physiologically flexibility, and therefore the less behavioral flexibility, the individual is thought to have to respond to environmental demands. i.e.: decreased self-regulation.

The Polyvagal theory further describes the evolution of the ANS and its relationship to social communication and behavior. The PVT proposes that the nervous system has evolved in such a way to allow for an individual to maintain homeostasis. This theory describes three phylogenic stages of development of the ANS which are ordered and behaviorally related to social communication and action. This includes the vagal regulation of visceral states to support social behavior. These three stages are: mobilization (fight or flight behaviors) which is under the control of the SNS and is the most primitive of all components, immobilization (behavioral shutdown, vaso-vagal syncope and feigning death) which is under the control of the unmyelinated vagus and social communication which is under the control of the myelinated vagus and is the most advanced system. These three systems are organized to respond to challenges in a phylogenetically ordered manner consistent with the Jacksonian principal of dissolution which states that in the brain, higher or newer neural circuits inhibit lower ones (Porges, 2007). Functionally, when the environment is perceived as safe, visceral homeostasis occurs via the influence of the unmyelinated portion of the vagus nerve and the components of the social engagement system are ready for action.

The Social Engagement System

Nested in the PVT is the notion of the social engagement system. The social engagement system is a set of physiologic processes that allow individuals to engage in pro-social and social behaviors. Pro-social behaviors include "determination of friend from foe and evaluation of the safety of the environment..... (Porges, 2005)". These primitive social behaviors (pro-social behaviors) begin with the evaluation of sensory input. Sensory information is the first way in which we evaluate the safety of a situation. This then determines how we proceed with our interactions. We react to the evaluation of sensory input through changes in physiologic state to meet the demands of the situation. These physiologic reactions then influence our behavioral responses.

The social engagement system further proposes that through evolution the brainstem nuclei that regulate the myelinated vagus became integrated with the nuclei that regulate the muscles of the face and head. This link resulted in a coupling between spontaneous social engagement and bodily state. The muscles of the face and head that regulate eye gaze and looking, facial expression, listening and prosody are neuroanatomically and neurophysiologically linked to visceral state. Specifically, the social engagement system has a control component in the cortex that regulates brainstem nuclei that control eyelid opening needed for looking and eye gaze, facial muscles needed for facial expression, middle ear muscles, specifically the stapedius muscle, which is central in extracting human voice from background noise, muscles of mastication which are involved in ingestion, laryngeal and pharyngeal muscles which play a major role in prosody and intonation of voice and the head turning muscles which allow for orienting and social gesturing. Collectively these pro-social and social functions act as filters to limit stimuli and allow us to engage within the social environment. Both the pro-social and social functions mentioned above appear to be deficient in persons with ASD.

The socially linked muscle groups (somatomotor) and the visceral components of the system are linked in that activation of one system can affect the other. For example activation of the somatomotor component of this system i.e.: looking, listening, head orienting etc… could trigger visceral states that would further support this engagement and allow the individual to continue with the interaction. The reverse is also true. Visceral changes that support social interaction i.e.: the myelinated vagal influence over the sino-atrial node of the heart, a.k.a the vagal brake, can support and foster social engagement behaviors such as looking, listening, and orienting.

In summary, the concept of the social engagement system proposes that the regulation of physiologic states, as controlled by the vagal system, mediates "the range of emotional expression, quality of communication and the ability to regulate bodily and behavioral state (Porges, 2005)". If one is not able to efficiently and functionally evaluate and respond, both physiologically and behaviorally, to the sensory information coming from the environment, and from one's own body, than one cannot possibly engage in meaningful social interactions because the physiologic conditions needed for social interactions are not initiated.

Given the theoretical perspectives of the polyvagal theory and the social engagement system it is reasonable to hypothesize that the regulatory sensory processing and social interaction difficulties characteristic of autism may be due in part to a functionally compromised vagal system. A potential theoretical framework which can be used to investigate this hypothesis is to monitor vagal tone in children with autism both at rest and during sensory challenges. At rest the vagal system should be at relatively high levels of activation in order to promote homeostasis and in the presence of a challenge vagal tone suppression should enable the individual to direct his/her attention outward and to meet the demands of the situation. Thus, the functional quality of the vagal system or vagal tone, can be assessed by measuring the nervous system's modulation of heart rate via the vagus nerve, a.k.a vagal tone.

Behavioral Responsivity to Sensation

Neurophysiologic measures alone however cannot give us a complete picture of an individual's regulatory-sensory processing abilities. Regulatory-sensory processing is a combination of internal processes coupled with behavioral responses. It is the combination of the neurophysiologic response with the behavioral response strategy that truly profiles an individual's regulatory-sensory processing abilities and adaptive functioning in the environment. There are a variety of possible combinations of neurophysiologic and behavioral response patterns to sensation that may explain some of the heterogeneity of symptoms characteristic of children with autistic spectrum disorders.

Dunn, (1999) describes a model of sensory processing (referred to as Regulatory-Sensory Processing in this paper) that accounts for the neurologic threshold for responding and the person's propensity for responding behaviorally to those thresholds. Neurologic threshold refers to the point at which a neuron or neuron system responds to information. When the threshold is met the physiologic changes necessary to meet the demands of the environment based on the evaluation of the incoming input are initiated. A high threshold indicates that a neuron needs a lot of stimulation to fire while a low threshold indicates that a neuron needs very little stimuli to fire. An individual's behavioral responses reflect their neurologic thresholds, they may act in accordance with their thresholds or attempt to counteract their thresholds. In this model thresholds and responding strategies are a continuum of possible conditions such that a person's way of responding to sensory events in daily life can be characterized as reflecting both a particular threshold and a responding strategy. Individuals that have high thresholds with passive responding strategies are categorized as having *poor registration*. These individuals tend to act in accordance with their threshold. They often appear disinterested with flat/dull affect and low energy/muscle tone because they are not receiving the adequate amount of input to meet their threshold and to initiate the physiologic changes needed for engagement and active attention. Individuals with high thresholds that have an active responding strategy are called *sensory seekers*. These individuals act in an attempt to counteract their threshold. They look to add additional sensation to everyday experiences in order to get more input from the experience. For instance they may make noises with their mouth while working, chew on things like pen caps or fidget in their chair in order to meet their threshold.

Individuals that have a low threshold with passive responding strategies fall into the category of *sensory sensitivity*. They act in accordance with their threshold. Individuals in the sensory sensitivity category may appear distractible and hyperactive because they are responding to every new sensation that enters their environment. On the other hand individuals with low thresholds that have an active responding strategy are called *sensory avoiding*. These individuals act to counteract their threshold. They very often have emotional outbursts or disruptive behaviors because the sensory information they are taking is uncomfortable or frightening. Further, they often appear rigid and controlling and may create rituals in an attempt to control the types and amount of sensation that enter their environment.

When viewed together the Polyvagal Theory and Dunn's model for sensory processing offer an explanation for many of the behaviors seen in individuals with autistic spectrum disorders. For example, if an individual has a low threshold for responding to sensory information, than that individual needs only a small amount of sensory information in order for the physiologic processes characteristic of vagal tone suppression to be initiated. This suppression may be characterized as poorly timed and poorly modulated to the nature or intensity of the situation. Depending on the individual, a passive (sensory sensitivity) or active (sensory avoiding) behavioral strategy will then be initiated in response to the physiologic changes. This low threshold pattern can be observed in many of the behaviors seen in individuals with autism such as sensory defensiveness, ritualistic behaviors (in an attempt to maintain consistency and control over the environment for fear of a stimuli entering that may be noxious) or the phenomenon of "shut down" where the individual withdraws to avoid external events that may be uncomfortable.

Similarly, a person with a high threshold may not appropriately suppress vagal tone appropriately in terms of intensity, duration or time frame. This results in a situation where the body is not prepared to direct attention outward to face the environmental demands. The individual will then use either a passive (poor registration) or active (sensory seeker) behavioral strategy to respond to this state of delayed or decreased intensity of vagal suppression. These high threshold patterns can also be seen in individuals with autism. Behaviors assumed to be resultant of high thresholds include stereotypic stimulating behaviors such as hand flapping, head shaking, banging or toe walking in an attempt to get more input (sensory seekers) as well as lethargy and unresponsiveness (poor registration).

Hence, the apparent heterogeneity of symptoms seen in persons with autistic spectrum disorders may be explained by a dysregulation of the vagal system resulting in combinations of high and low thresholds and individual differences in responding to those thresholds.

The Sensory Profile

The *Sensory Profile* (Dunn, 1999) is a behavioral measure of sensory processing. The Sensory Profile is a widely used caregiver questionnaire that provides a method for professionals to measure a child's sensory processing abilities and to profile the effect of sensory processing on functional performance. The questionnaire is grouped into three main scoring sections: Sensory Processing, Modulation and Behavioral and Emotional Responses. The Sensory Processing section indicates the child's responses to basic sensory information. This section is further divided into 6 categories Auditory, Visual, Vestibular, Touch, Multisensory and Oral Sensory Processing. The Modulation section reflects the child's neural and behavioral modulation of sensory information. This section is further divided into 5 subsections, Sensory Processing Related Endurance/Tone, Modulation Related to Body Position and Movement, Modulation of Movement Affecting Activity Level, Modulation of Sensory Input Affecting Emotional Responses, and Modulation of Visual Input Affecting Emotional Responses and Activity Level. The Behavioral and Emotional Responses section reflects the child's behavioral outcomes of sensory processing. This section is further divided into 3 sections, Emotional/Social Responses, Behavioral Outcomes of Sensory Processing and Items Indicating Thresholds for Response.

The items on the Sensory Profile are further categorized into 9 factorsbased on a factor analysis conducted by Dunn and Brown (1997). Factor1-Sensory Seeking, Factor 2-Emotionally Reactive, Factor3-Low Endurance/Tone, Factor4-Oral Sensory Sensitivity, Factor5-Inattention/Distractibility, Factor6-Poor Registration, Factor7-Sensory Sensitivity, Factor8-

Sedentary, and Factor9-Fine Motor/Perceptual. Item ratings are based on a five point likert scale; 1= Always, 2= Frequently, 3= Occasionally, 4= Seldom and 5= Never for each question.

All scoring is based on normative data from the performance of typically developing children (n=1,037) (Dunn & Westman, 1997). The professional scoring the questionnaire adds up the raw score for each sub-section to obtain a section raw score based on normative data. The sub-sections raw scores are then plotted in the section summary table to classify the section raw score totals into one of three categories, Typical Performance, Probable Difference and Definite Difference. Typical Performance is scores at or above point1 standard deviation (SD) below the mean (M). This range indicates that the child performed similar to a child in the top 84% of the children studied without disabilities. Probable Difference is scores at or above 2 standard deviations (SD) below the mean. This range of scores indicates that the child's performance was between the 2^{nd} and 16^{th} percentile, representing 14% of the sample of children without disabilities. Definite Difference is scores 2 SD below the mean. These scores indicate that the child is performing similar to the lowest 2% of children without disabilities. The lower the overall raw score the greater the dysfunction. The item raw scores are then plotted into the factor groupings to obtain a factor raw score. These factor raw scores are then plotted in the Factor Raw Score Summary which classifies the child's behavior using the same three categories: Typical Performance, Probable Difference and Definite Difference. These classifications are also based on the performance of children without disabilities.

The Sensory Profile is based on W. Dunn's model of sensory processing described above which may serve as a guide to help us re-frame the apparent heterogeneous behavioral symptoms seen so often in children with sensory modulation disruptions including children with autistic spectrum disorders. However, while it is important to profile behaviors in order give us a better understanding of a child's sensory processing patterns this alone cannot provide a complete picture of an individual's regulatory sensory processing abilities. As mentioned above, regulatory-sensory processing is a combination of internal processes coupled with behavioral responses. It is the combination of the neurophysiologic response with the behavioral response strategy that truly profiles an individual's regulatory-sensory processing abilities and adaptive functioning in the environment. Understanding the physiologic patterns of sensory processing may contribute to the understanding of the behaviors in groups of children with regulatory sensory processing disorders including children with autistic spectrum disorders.

In summary, when viewed in the framework of neuro-occupation, the Polyvagal Theory provides a theoretical perspective that explains the role of the vagal system in disorders of regulatory sensory processing, including children with autistic spectrum disorders. It can be hypothesized that children with ASD have difficulties with regulatory sensory processing as a result of autonomic dysregulation and that these difficulties result in physiologic inflexibility in responding to environmental demands. Investigation into the neurophysiologic functioning of children with autism in response to sensation may provide valuable information on the physiologic underpinnings of the behavioral response patterns seen in this population and the behavioral symptomology encountered. However, it is first necessary to determine the reliability of the measures of sensory processing being used in order to determine the validity of the results of such studies, as well as to lay a foundation for future studies of treatment approaches aimed at reducing abnormal behavioral responsivity to sensation. Hence, the purposes of this study are to examine three related research questions: 1) Is cardiac vagal tone a reliable measure of regulatory sensory processing in typically developing children and children with autistic spectrum disorders? 2) Do children with autistic spectrum disorders respond to sensation (behaviorally and physiologically) differently than typically developing children? 3) Is there a relationship between physiologic and behavioral responses to sensation in children with and without autistic spectrum disorders?

Chapter II

Literature Review

This literature review will focus on the research involving vagal tone as it relates to regulatory –sensory processing. However, as discussed in the introduction to this paper, the diagnoses of sensory processing disorder and regulatory disorder have been recently retermed regulatory -sensory processing disorder. As such, any literature published before mid-2005 will refer to each of these diagnoses separately.

The information presented is divided into four sub-sections for ease of reading. The first sub-section reviews the literature on the on baseline vagal tone and it's relationship to self-regulation and sensory modulation, collectively known as regulatory sensory processing, and development. The second sub-section reviews the information on vagal regulation as it relates to self- regulation, including sensory modulation ,collectively known as regulatory sensory processing, and attention and social interaction in children. The third sub-section reviews the literature describing the relationship between baseline vagal tone and the regulation of vagal tone in children. The fourth sub-section focuses on a review of the literature of the reliability of vagal tone in children. Finally, a review of the literature on the Sensory Profile involving children with autism is also featured in this review.

It should be noted that much of the research done in this area involves infants and young children. There are very few studies that look at school aged children. In addition, there are currently no vagal tone studies available in the literature involving children with autism spectrum disorders.

Baseline Vagal Tone

Baseline vagal tone refers to an individual's vagal state at rest. This state is thought to be an indicator of overall physiologic stability and has been linked to self -regulatory abilities. This section of the literature review will focus on the research that supports the relationship between baseline vagal tone and self-regulation in infants and children.

The role of baseline vagal tone in self-regulation and developmental outcomes in infants has been widely investigated (Porges et al, 1996; Fox, 1989; DeGangi et al, 1991; Fox and Porges, 1985; Huffman, L.C., Bryan, Y.E., delCarmen, R., Pedersen, F.A., Doussard-Roosevelt, J.A. & Porges, S.W., 1998; Gunnar, M.R., Porter, F.L., Wolf, C.M., Rigatuso, J. & Larson, M.C., 1995; Stifer and Fox, 1990; Richards, 1985). Huffman et al. (1998) found that three-month-old infants with high baseline vagal tone demonstrated fewer negative behaviors and required less calming and soothing. Fox (1989) and Stifter and Fox (1990) found that five-month-old infants with high baseline vagal tone as compared to infants with low baseline vagal tone, were more appropriately reactive to both positive and negative events. These same infants when tested at fourteen months demonstrated a greater exploration and awareness of environment and attentional capacities.

In a longitudinal study, Porges et al. (1996) measured baseline vagal tone in infants at nine months and at three years and had their mothers complete the Child Behavior Checklist (CBCL). Infants who at nine months had low baseline vagal tone demonstrated more problems on the following subscales of the CBCL – sleep problems, somatic behavior, depressive and destructive behavior, all areas which are indices of regulatory-sensory processing abilities. In another longitudinal study, Fox and Porges (1985) studied the association of vagal tone and developmental outcomes at 8 and 12 months of age using the Bayley Scales of Mental

Development (BSMD). Four groups of children were studied: premature infants without medical complications; premature infants with respiratory distress syndrome (RSD); term infants with birth asphyxia; healthy term infants. Three minutes of resting vagal tone was analyzed on each infant in the hospital. The results showed that infants with high vagal tone always had high BSMD scores at 8 and 12 months while infants with low vagal tone presented with varied outcomes.

In addition to studies of infants, baseline vagal tone has also been examined in older children with suspected disruptions in regulatory-sensory processing. Boccia and Roberts (2000) found that boys with Fragile X Syndrome, who are hypersensitive to stimuli, also had decreased vagal tone at rest. Schaaf, Miller, Seawell and O'Keefe (2003) demonstrated that children with sensory modulation disruption (SMD) had lower baseline vagal tone than children without SMD. Schaaf et al (2010) found that children with severe SMD, determined by falling four standard deviations below the mean on multiple sections of the Short Sensory Profile (Dunn, 1991), were more likely to demonstrate lower vagal tone at baseline as well as during auditory stimulation during the administration of the Sensory Challenge Protocol (SCP), a laboratory paradigm designed to measure physiologic responsivity to sensation.

Together these studies suggest that infants with higher resting baseline vagal tone demonstrate better self-regulatory abilities and better developmental outcomes. In addition, the results of two studies demonstrate that older children with suspected disruptions in sensory processing i.e.: children with fragile x syndrome and primary sensory modulation disruption (SMD), have lower baseline vagal tone than typically developing peers. These findings are an important first step in identifying a link between behavioral manifestations and neurophysiologic processes. To date, there have not been any studies looking at baseline vagal tone in children with autistic spectrum disorders. Investigation into the baseline vagal functioning of children with autistic spectrum disorders may provide us with valuable information regarding the regulatory-sensory processing abilities and developmental issues often encountered in this population.

Regulation of Vagal Tone

Vagal regulation, specifically, suppression of vagal tone, has been related to social interaction skills and attentional capacities, both functions of efficient regulatory-sensory processing. The suppression of vagal tone during a challenge or event is considered a mechanism for directing attention outward. This skill is essential for both social interactions and achieving and maintaining attention. Several studies have investigated the relationship between suppression of vagal tone with a challenge and social interaction and attentional capabilities (Porges et al, 1996; Huffman et al, 1998; Fox, 1989; Calkins, 1997; Suess, P.E., Porges, S.W. & Plude, D.J, 1994; DeGangi et al., 1991;Gunnar et al., 1995; Stifter and Fox.,1990; Richards, 1987).

Porges et al. (1996) measured the amount of vagal suppression in 9-month-old infants during the administration of the Bayley Scales of Mental Development (BCMD). A significant correlation was found between vagal tone suppression at 9 months and scores on the Child Behavior Checklist (CBCL) at three years. The greater the suppression of vagal tone at 9 months, the more positive the outcomes on the three subscales of the Child Behavior Checklist (CBCL) – social withdrawal, depressed and aggressive. Huffman et al. (1998) studied infants at three months of age using a laboratory paradigm called the Behavioral Response Paradigm, which consists of 30-second episodes of social interactions or sensory stimulation. Subjects were rated on positive or negative reactivity, sociability, soothing or calming required and overall activity.

They found that infants who consistently suppressed vagal tone during these events and had larger drops in vagal tone ,demonstrated more prosocial and attentive behaviors. In contrast, infants who did not consistently suppress vagal tone during the event were rated as more difficult to sooth and had shorter periods of orienting. Gunnar et al. (1995) measured vagal tone in 50 newborns during a heel stick procedure. They found that greater vagal tone suppression during the heel stick procedure (a stressful event) was related to later reports (6 months) of less negative infant temperament on an infant temperament scale. Calkins (1997) studied the vagal reactions of 2-3 year old children during a four-episode sequence of events designed to elicit positive and negative reactions, attention, and social interaction. She found that the greater the suppression of vagal tone during each event the more the child looked at and interacted with the experimenter. The children who demonstrated less vagal tone suppression had more self-orienting behaviors (withdrawn) and delayed task orientation (slowed processing/initiation).

In contrast, Suess and Bornnnstein (2001), found that suppression of vagal tone in 20 month old children occurred with cognitive tasks only. During social tasks vagal re-engagement, i.e. increases from a suppressed state of vagal tone during a cognitive task, were reported. And in fact, this re-engagement predicted the language and play performance of the children on those specific social tasks, i.e.: children with larger increases had better language and play performance. Schaaf et al, (2010) examined vagal regulation in typically developing children and children with Sensory Modulation Disorder (SMD) ages 5-12 years, during the administration of the Sensory Challenge Protocol (SCP). The SCP is a laboratory paradigm designed to measure physiologic responsivity to sensation. Five domains of sensation are measured, auditory (a 78 decibel siren), visual (a 20 watt strobe light at 10 Hz), olfactory (winter green oil), tactile (a feather lightly moved along the face) and vestibular (chair tilted back to a 30 degree angle), with eight consecutive trials of each lasting three seconds per trial. There is a 12-17 second time period between each trial and a 20-30 second time period between each sensory domain. There was also a prolonged (2 minute) auditory stimulus (tone) that was presented after the recovery period. Baseline (resting) and recovery (return to baseline) periods were also recorded. They found that typically developing children demonstrated minimal reactivity from baseline to each sensation and had change scores close to zero. Whereas children with SMD demonstrated an increase in vagal tone from baseline to each of the sensory domains with the exception of tones and olfaction.

Hence, much of the research on vagal regulation in infants and children suggest that children who consistently suppress vagal tone and who demonstrate greater drops in vagal tone in response to an event demonstrate overall better attention and social interaction skills and thus more efficient regulatory-sensory processing. However, there is also contradictory information demonstrating minimal reactivity or a rise in vagal tone with a challenge in both typically developing children and children with SMD (Suess and Bornstein (2000); Schaaf et al, (2010)). To date no studies have been published looking at the relationship between vagal regulation and regulatory-sensory processing in children with autistic spectrum disorders and there has been very few studies looking at the relationship between vagal regulation and regulatory-sensory processing in typically developing school aged children. Understanding the vagal responsivity to sensation of school aged typically developing children may provide a foundation upon which we can compare vagal responsivity to sensation of school aged children with autistic spectrum disorders to sensation.

The Relationship between Baseline Vagal Tone and Regulation of Vagal Tone

The relationship between baseline vagal tone and vagal regulation has also been investigated. Degangi, G.A et al. (1991) measured both baseline vagal tone and vagal regulation in 8-11 months old babies with and without regulatory disorders during the administration of the Bayley Scales of Mental Development and the Test of Sensory Functions in Infants. Typically developing babies with high baseline vagal tone consistently suppressed vagal tone during the information processing tasks on the tests. However, the babies with regulatory disorders who demonstrated high baseline vagal tone did not consistently suppress vagal tone with information processing. Porges et al.'s (1996) work supports these findings. They found that infants who had higher baseline vagal tone also demonstrated greater suppression of vagal tone during the administration of the Bayley Scales of Mental Development. These same infants at a three-year follow up had more optimal scores on the social interaction subtests of the CBCL and fewer behavioral problems. Calkins (1997) also found a relationship between baseline vagal tone and vagal tone suppression in children. Children 2-3 years old with higher baseline vagal tone consistently suppressed vagal tone during and event or challenge. Children with low baseline vagal tone inconsistently suppressed or did not suppress at all. These findings suggest that there may be problem in the balance between baseline vagal tone and vagal regulation in response to a challenge in babies and young children with regulatory sensory processing disorders.

Suess et al. (1994) measured both baseline vagal tone and vagal regulation in fourth and fifth grade students, mean age 9.10 years, during a continuous performance task (CPT). The task required the children to press the spacebar on the computer each time the number 5 appeared after the number 1 within the target array on the computer screen. The total task time was 9 minutes. There was no relationship found between baseline vagal tone and vagal tone regulation.

All children tested, including those with high baseline vagal tone and those with lower baseline vagal tone, demonstrated a decrease in vagal tone during the CPT. These results may be explained by the fact that older children, through experience, learn the self-regulatory abilities needed for suppression of vagal tone during situations in which sustained attention is required. Infants and young children on the other hand have not yet learned this skill since self-regulatory abilities (physiologic and behavioral) are thought to increase with maturation. Future studies of vagal tone regulation in older typically developing children and children with attentional and regulatory –sensory processing disorders are needed to test this hypothesis.

Based on the above mentioned studies, there appears to be a functional relationship between baseline vagal tone and vagal regulation in response to an event. Infants and young children with high baseline vagal tone more consistently suppress vagal tone in response to a challenge. This relationship indicates the supportive function of baseline vagal tone in adapting to environmental demands. Currently there is no conclusive information on whether or not this same pattern holds true in older children with and without disabilities including children with autistic spectrum disorders. Thus, future studies with these populations are needed to clarify the relationship between baseline vagal tone and vagal regulation.

Reliability of Vagal Tone

While there is a fair amount of research published using vagal tone as a measurement of self- regulation in typically developing infants, young children and some school aged children, there is a limited amount of information on the reliability of vagal tone as a measure with these populations. And while there is also a fair amount of information on infants with regulatory disorders there is *NO* information on the reliability of the measurement in this population.

Finally, vagal tone has been used and continues to be used to asses physiologic responsivity during the administration of the Sensory Challenge Protocol as described above, in children with various regulatory sensory processing disorders including SMD and ASD. However, there is no information on the reliability of this measure with these populations. Without reliability information the strength of the findings from previous studies is limited.

As mentioned above, the reliability information on vagal tone as a measure of regulatory-sensory processing in infants and children is limited and comparing the results from the studies is difficult for several reasons: first, because the infants and children in these studies are of varying ages; secondly, because some studies looked at long term reliability across years while other studies examined short term reliability across days or weeks; lastly because various types of tasks were used to measure vagal regulation.

The reliability of baseline vagal tone measures in infants and children of various ages and over varied time periods has been investigated (Stifter and Fox, 1990; Porges, 1992; Calkins and Keane, 2004; El-Sheik, 2005; Doussard- Roosevelt et al., 2003). Many of these studies involve only typically developing children and have differing results. Porges (1992) measured resting vagal tone of 16 infants in a neonatal intensive care unit for 5 consecutive days. He found that baseline vagal tone remained consistent and stable over these five days. Stifter and Fox (1990) however measured resting vagal tone in typically developing infants in the nursery and then measured resting vagal tone again at 5 months of age and found that vagal tone did not remain consistent. Infants at 5 months demonstrated lower baseline tone than they did as newborns. Calkins and Keane (2004) found that there was only modest stability of baseline vagal tone across time in children tested at 2 years and then 4.5 years. Finally, El-Sheik (2005) examined baseline vagal tone in children at 9 years and again at 11 years of age and found that baseline vagal tone remained stable and consistent from 9 to 11 years of age.

The reliability of vagal regulation has also been minimally studied (Doussard- Roosevelt et al., 2003; Calkins and Keane, 2004; El-Sheik, 2005). Doussard –Roosevelt et al. (2003), measured the vagal regulation of typically developing kindergarten children to a negative-affect elicitor task over a four week period. They found modest stability over the first two week period with no stability over the second two week period. However, they did use the same task each of the four weeks therefore habituation to the task may have occurred by week three which may explain the inconsistency. Calkins and Keane (2004), in a longitudinal study, measured vagal regulation of 125 typically developing children in response to 5 different tasks that required different emotional and attentional demands. The tasks included: baseline, attention, empathy, frustration, problem solving. Children were tested at age 2 years and then again at 4.5 years. Although there were differences in responses between tasks, there was modest stability of vagal tone overall from 2.0 to 4.5 years. Lastly, El-Sheik (2005), tested typically developing children at 9 years of age and then again at age 11years on a cognitive (star tracing from a mirror image) and emotional task (listening to adults argue). He found stability over time in vagal tone response to the cognitive task but not for the emotional task (vagal suppression to the emotional task was less at 11 years). It can be speculated that over the two year testing period children developed emotional maturity and therefore the physiologic response to an emotionally driven task was not as great.

Based upon the data from studies on typically developing children it appears as though the reliability of vagal tone may be dependent on age and maturational factors as well as by the type of task used to elicit a vagal regulatory response. More information is needed to identify specific patterns of vagal responsivity and it's stability over time in typically developing children. In addition, the reliability of vagal tone measurements in children who are not typically developing is still questionable and needs to be examined.

Summary of Literature Review of Vagal Tone

Several themes can be extracted from the plethora of research on vagal tone in children. First, infants with high resting baseline vagal tone demonstrate greater regulatory-sensory processing abilities and greater developmental outcomes. The second theme is that older children with suspected disruptions in regulatory-sensory processing i.e.: children with fragile x and sensory modulation disruption (SMD) have lower baseline vagal tone than typically developing peers. More studies are needed to determine if this holds true with other groups of children with regulatory sensory processing disorders including children with autism spectrum disorders. The third theme evident in the literature is that children who consistently suppress vagal tone in response to an event demonstrate overall better attention and social interaction skills. Fourth, there appears to be a functional relationship between baseline vagal tone and suppression of vagal tone in response to an event. Infants and young children with high baseline vagal tone more consistently suppress vagal tone in response to a challenge. It is unclear whether this same relationship holds true in older children with and without disabilities. Finally, it appears as though the reliability of vagal tone may be dependent on age and maturational factors as well as by the type of task used to elicit a vagal regulatory response. More information on the reliability of baseline vagal tone and vagal regulation in both typically developing children and children in various diagnostic groups is needed.

Measures of vagal tone can provide insight into the neurophysiologic mechanisms underlying regulatory-sensory processing, attention and social interaction, all of which are deficient in children with autistic spectrum disorders. More information however, is needed on the reliability of the measure in both typically developing children and children with various diagnoses including autism, in order to validate and strengthen previous findings and determine the use of this measure for use over time.

The Sensory Profile

Measuring physiologic responses alone does not provide us with a complete picture of an individual's regulatory-sensory processing abilities. It is the combination of the neurophysiologic responses with the behavioral response strategies that truly profiles an individual's regulatory-sensory processing abilities and adaptive functioning within the environment.

The Sensory Profile is a behavioral measure of sensory processing that has been validated for use in children with autistic spectrum disorders (Ermer and Dunn, 1998; Kietz and Dunn, 1996; Watling, R., Dietz, J. & White, O., 2000). Dunn and Kientz (1996) compared the scores of children with and without autism on the Sensory Profile. Children with autism were also given the Childhood Autism Rating Scale (CARS) to determine the severity of the disorder. According to the CARS fifteen children with autism were classified as being mild/moderately autistic and seventeen severely autistic. There were no significant differences on Sensory Profile scores between the subjects with mild/moderate autism versus subjects with severe autism in any category and therefore the groups were collapsed for further analysis. Univariate analysis demonstrated that 85% of the items contributed to the differences between groups. In fact, items that were most representative of children with autism were least common for children without autism. However, no items on the Sensory Profile met the criterion for being "common" in children with autism (80% or more reporting by the parent that the child displays a certain behavior). This finding is not surprising considering the heterogeneity of children with autistic spectrum disorders. However, there were certain items that parents reported observing in their children 50% of the time. These items were scattered throughout the categories except for the Social/Emotional category in which more than half of the behaviors were reported to occur in children with autism.

Watling, Deitz and White (2000) compared Sensory Profile scores of children ages 3-6 years with and without autism. The results confirmed what Dunn and Kientz (1996) reported; children with autism have scores that vary widely and are significantly different, and overall lower, than children without autism. For example children with autism displayed a greater frequency of the behaviors on the Emotionally Reactive factor than children without autism (67.5%); on the Low Endurance/Tone factor children with autism received scores across a broad range whereas children without autism clustered at the highest end of the scale; on the Poor Registration factor 62.5% of children without autism scored higher than children with autism; and on the Other factor 65% of the children with autism had lower scores than children without autism. Furthermore, the children with autism scored lower on a greater number of factors than children without autism. Specifically, children with autism scored lower than children without autism on a range of 0-6 factors, with 34 children (85%) receiving scores lower than any of the children without autism on at least 1 factor. Of those 34 children, 6 had scores lower than any child without autism on 1 factor, 11 on 2 factors, 6 on 4 factors, 4 on 5 factors and 2 on 6 factors.

The literature on behavioral responsivity indicates that children with autism demonstrate behavioral response patterns that are significantly different than children without autism. Again, the question then arises as to what neurophysiologic processes may be underlying these behavioral responses? However, before we can answer this question we need to determine the reliability of the measurements used to index these neurophysiologic processes.

Study Purpose

The purpose of this study is to examine three related research questions: First "Is cardiac vagal tone a reliable measure of regulatory sensory processing in typically developing children and children with autistic spectrum disorders?" ; Second, "Is there a relationship between physiologic and behavioral responses to sensation in children with and without autistic spectrum disorders?" ; Lastly, "Do children with autistic spectrum disorders respond to sensation (behaviorally and physiologically) differently than typically developing children?". These questions were explored by measuring physiologic responsivity to sensation, via cardiac vagal tone, two times over a three-week period and behavioral responsivity to sensation via The Short Sensory Profile, in boys with and without autism. Physiologic and behavioral measures for the two groups were compared, as was cardiac vagal tone measures per individual and per group for each testing session.

Chapter III
Methods

Design

A multi-factorial repeated measures design was used to examine the reliability of vagal tone as a measure of regulatory sensory processing and to investigate the relationship between physiologic and behavioral measures of sensory processing, in children with and without autistic spectrum disorders. The independent variables were group, sensation and testing session. The dependent variables were physiologic and behavioral responses to sensation. The experimental group was boys with autistic spectrum disorders ages four to eleven years. The control group was comprised of typically developing boys age's four to eleven. The physiologic measure that was analyzed in this study was vagal tone and the behavioral measure was the Short Sensory Profile (Dunn, 1997).

The Seton Hall Institutional Review Board (IRB) approved this research proposal in April 2006. A copy of this approval letter is found in Appendix B.

Participants

A total of thirty nine boys with and without autism were tested (n=16 controls and 23 with ASD). Participants were recruited from specialized educational programs and therapy clinics in New Jersey via letters of solicitation and flyers. See Appendix C and D for solicitation letter and flyer. Flyers were also posted in common areas of Seton Hall University asking for volunteers.

Parents interested in finding out more information on the study contacted the primary investigator via phone or email. When parents contacted the investigator they were asked routine questions about their child's medical history such as when their child was given the diagnosis of autism (if talking to a parent of a child with autism), whether their child has any existing (if talking with a parent of a typically developing child) or co-existing diagnosis (if talking with the parent of a child with autism) including hearing or vision loss or any medications that child may be taking. See Appendix E for copy of medical history form. Any child with medical conditions aside from autism was excluded from the study to rule out the possibility that the condition might affect the child's responsivity to sensation during the testing. Children who had sensory hearing loss or visual impairments were excluded from the study since the study was looking at responses to auditory and visual information. Children who were taking any medication that is known to affect arousal were excluded from this study for the same reason. Parents were asked simple questions about their child's receptive language skills and activity level. Children who were unable to follow a simple command were excluded from the study.

Since it is unknown whether males and females process sensory information in the same manner, we limited the subjects to males since the majority of children with ASD are boys and gender differences in the patterns of symptoms between the two genders may be present (Lord, Schopler and Revicki, 1982).

The average age for the control group was 8.7 (SD=1.51) years and for the experimental group was 7.3 (SD=1.18) years.

Measures to ensure confidentiality

Any forms that contained the names of participants were kept in a locked cabinet that was accessed only by researchers. All collected data was assigned a coding system and reported in aggregate form. Information collected during the course of this study will be kept for 3 years and then shredded.

Instrumentation

In order to maintain consistency with the labs run by the researchers in the SIRC we chose to use the same instrumentation to allow for comparing results across sites. Heart rate was measured during all testing sessions using the Psylab SAM acquisition and analysis system by Contact Precision Instruments®. The Psylab SAM is an integrated laboratory system that allows for simultaneous collection of multiple forms of physiologic data including heart rate and skin conductance. The data was graphically displayed on a computer monitor as it was collected and was later converted to numeric values for analysis. The Psylab SAM system also allowed for synchronization of external and physiologic events.

Skin conductance was recorded simultaneously with heart rate for future analysis separate from this dissertation.

Behavioral responsivity to sensation was measured using the Short Sensory Profile (Dunn, 1997). The Sensory Profile is a caregiver questionnaire that measures behavioral responsivity to sensation. The Sensory Profile has been validated for use in children with and without autistic spectrum disorders (Ermer and Dunn, 1998; Kietz and Dunn, 1996; Watling, R., Dietz, J. & White, O., 2000). The Short Sensory Profile is a condensed version of the Sensory Profile intended for use in research.

Procedures

Testing occurred twice over a one month period at one of our testing sites. The testing environment was a quiet, dimly lit room with minimal distraction. Parents were encouraged to remain in the room with their children during the testing. Researchers acquainted both parent and child to the testing environment. The researcher explained the procedures involved in the experiment using age appropriate and lay terms. If the subjects and their parents agreed to participate in the study they signed the appropriate consent/assent form. Copies of consent and assent forms are found in Appendix F and G respectively.

Data Collection

At the initial testing session parents were asked to complete the Short Sensory Profile describing their child's behavioral responses to sensation. The parents were also asked to provide the researcher with identifying information which included date of birth and intervention history. During this time the researcher introduced the child to the testing environment by showing him the equipment and explaining the procedures to him in age appropriate language. When the child appeared to be comfortable in the setting, the researcher asked the child to sit in a sturdy armchair while electrodes were placed in a straight line directly below the rib cage. Smaller hand electrodes were placed on the child's thenar and hypothenar eminences on his left hand. All electrodes used hypoallergenic adhesive.

Heart rate and skin conductance were recorded for a two-minute baseline period during which the child was asked to remain still and quiet at both testing sessions . Heart rate and electrodermal reactivity were also recorded at all testing sessions as the Sensory Challenge

Protocol was administered (Miller, 1999). The Sensory Challenge Protocol (SCP) is a laboratory paradigm designed to measure physiologic responsivity to sensation in children and is the paradigm being used by researchers in the SIRC. Once again, we chose to use the same protocol in order to compare results across sites. Five domains of sensation were measured, vestibular (chair tilted back to a 30 degree angle), tactile (a feather lightly moved along the face), auditory (a 90 decibel siren), visual (a 20 watt strobe light at 10 Hz) and olfactory (winter green oil), with ten consecutive trials of each lasting three seconds per trial. There is a 15-19 second time period between each trial and a 20 second time period between each sensory domain. The testing was paused and breaks were given to the child as necessary. If at any time the child indicated, verbally or gesturally, that he/she wished to stop or if the child was in apparent distress, the experimenter stopped the current stimulus immediately. A sticker reward system was occasionally used when testing children with autistic spectrum disorder to encourage participation throughout the protocol. At the end of the testing session the experimenter thanked the child and parent/guardian and the child chose a small gift.

Data Analysis

Based upon power estimates from a previous pilot study using the same equipment and protocol but with a different population (Schaaf, R.C., Miller, L.J., Seawell, D., O'Keefe, S., 2003), a sample size of twenty subjects per group was determined as necessary, with an effect size of .90 and alpha set at .01.

A total of thirty nine children were tested. Several files could not be analyzed due to excessive artifact (n= 4 subjects with autism) or technical difficulties (n=2 subjects with ASD and n= 1 control). Further, several testing sessions had to be terminated due to inability of the subject to tolerate the protocol (n=6 subjects with autism). Thus, data files from twenty six boys, eleven with autism and fifteen typically developing, were included in this analysis.

Heart rate data was transferred from the Psylab to the MxEdit software analysis program (Porges, 1985) for artifact editing and quantification of the vagal tone index, which extracts the parasympathetic influence on heart rate via the vagus nerve. Heart rate was analyzed by the Porges (1985) method of calculating the respiratory sinus arrhythmia (RSA). This method applies an algorithm to the sequential heart rate data that identifies periods in heart rate slower than RSA. A band pass filter was then used to extract the variance of the heart rate periods within the frequency band of spontaneous respiration which for children is estimated .24-1.04 Hz. The average RSA was derived by calculating the natural log of this variance and is reported in ln(ms)2. The RSA was calculated continuously every 30 seconds during the SCP and a mean RSA was calculated for each domain.

Short Sensory Profiles were all scored by hand by the primary investigator or trained research assistant according to scoring procedures found in the Sensory Profile Manual (Dunn, 1999).

The first research question " Is cardiac vagal tone a reliable measure of regulatory sensory processing in typically developing children and children with autistic spectrum disorders?" was analyzed using and interclass correlation coefficients (ICC) to examine the stability of baseline measures and the stability of vagal tone changes in response to sensation between the two sessions.

The second research question " Do children with autistic spectrum disorders respond to sensation (behaviorally and physiologically) differently than typically developing children?" was analyzed first by performing an independent t-test on baseline scores to look for between group differences. This was done to determine whether baseline should be considered a covariate. Next a multivariate analysis of variance (ANOVA) was performed to examine between group differences on each domain of the SCP and pairwise comparisons using Bonferroni method were used to detect specific between group differences. Finally, a repeated measures analysis of variance was conducted to examine within group effects in vagal responses on the domains of the sensory challenge protocol. The assumption of sphrecity was checked using Mauchly's test. Newman Keuls post hoc test was performed to look for specific differences in vagal responses on SCP domains between groups. A multivariate analysis of variance was performed to examine between group differences on the SSP and pairwise comparisons using Bonferroni method were used to detect specific between group differences.

The third research question " Is there a relationship between physiologic and behavioral responses to sensation in children with autistic spectrum disorders and typically developing children?" was examined using Pearson Product Moment correlation coefficients to determine the relationships between vagal tone and Sensory Profile scores for both groups. Any data that did not meet the assumptions of normality were analyzed using the Spearman Rho Correlation Coefficient.

Average response scores (the average score across the sensory domains (tones, visual, sirens, tactile, olfactory and vestibular) of the SCP were calculated for each subject for both testing sessions and were analyzed in the same way as the domains of the SCP.

Change scores (difference from baseline to domain) were also calculated to control for baseline differences during between group non parametric procedures.

Chapter IV

Results

Data from the recovery 2 domain of the SCP, (the recovery domain of the second testing session) for the ASD group, was found not to be normally distributed based on the results of the Shapiro-Wilk test of normality. As such, nonparametric statistics were used for the analysis of the data from this domain.

Reliability

Reliability of the total subject pool was high with ICC's ranging between .782-.920. Reliability of the data from the typically developing group ranged from high to moderate reliability ranging between .525 - .923.

In contrast, the group of children with ASD demonstrated high reliability for all measures ranging between .776-.916. See Table 1 for ICC values.

Table 1. Test Re-test Reliability Measures of Vagal Tone for the Sensory Challenge Protocol.

SCP Domain	Total			Typical			ASD		
	M	SD	**ICC**	M	SD	**ICC**	M	SD	**ICC**
Baseline	6.36	1.20	**.830**	6.80	.981	**.702**	5.77	1.27	**.889**
Tones	6.57	1.16	**.782**	7.0	.933	**.525**	6.01	1.25	**.905**
Visual	6.59	1.05	**.830**	6.91	.847	**.650**	6.11	1.71	**.911**
Sirens	6.59	1.01	**.828**	7.01	.775	**.724**	6.03	1.07	**.819**
Olfactory	6.42	1.24	**.920**	6.91	.929	**.923**	5.74	1.33	**.881**
Tactile	6.55	1.17	**.912**	7.0	.779	**.810**	5.85	1.27	**.916**
Vestibular	6.76	1.11	**.916**	7.31	.711	**.837**	6.01	1.13	**.896**
Recovery	6.42	1.17	**.851**	6.90	.899	**.710**	5.77	1.21	**.899**
Av. Response	6.57	1.11	**.894**	7.04	.865	**.801**	5.94	1.18	**.901**

*n= 26

Within group between session differences

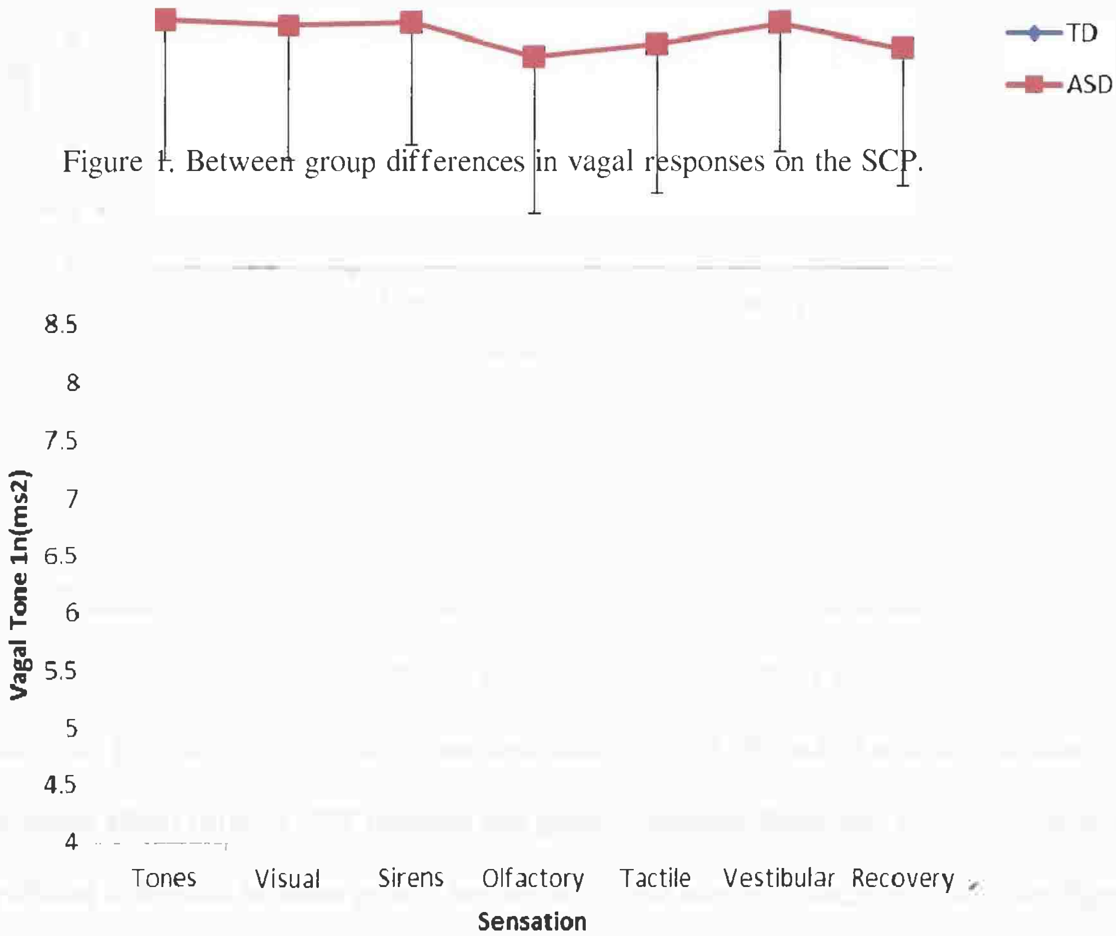
Within group between session differences were analyzed using repeated measures analysis of variance except for data from the recovery domain in the group of children with ASD. Since data from this condition was not normally distributed, between session differences were analyzed using the Wilcoxon signed-rank test. The assumption of sphericity was checked using Mauchly's test, and the Bonferroni method was used to perform pairwise comparisons. There were no significant differences in between session pairwise comparisons for either the control or experimental group. Since no significant differences were found between sessions for either group, the data was collapsed and all further analysis was conducted using the collapsed data.

Within group differences between baseline and recovery were examined using a paired t – test for the typically developing group and a Wilcoxon signed ranked test for the ASD group. There was no significant difference between baseline and recovery for the typically developing group, $t(14) = -.961$, $p=.353$, nor the ASD group, $p=.306$.

Between group differences on vagal measures

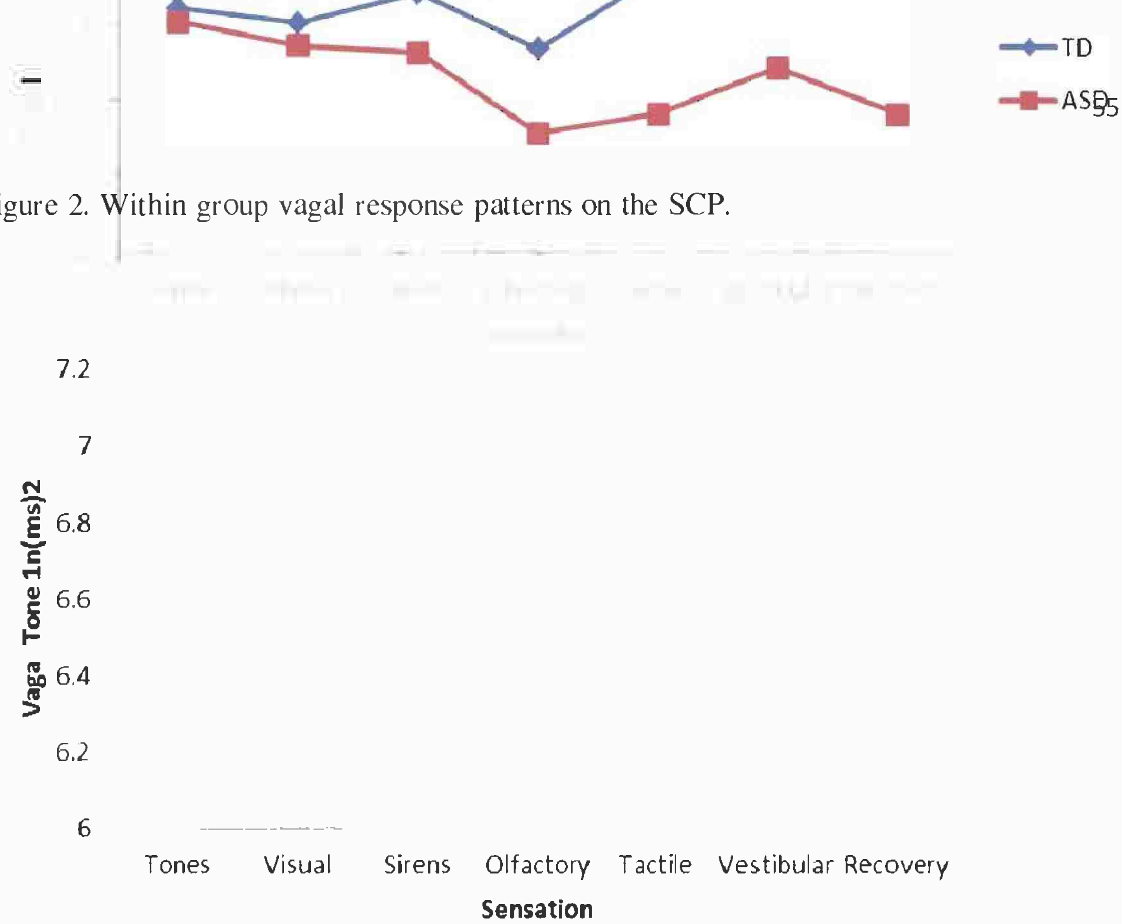
A t - test performed on baseline measures indicated that there was a significant difference, $t(20)=2.58$, $p=.018$, between the typically developing group (M=6.80, SD=.981) and the ASD group (M= 5.77, SD=1.27). As such, baseline was used as a covariate in both the multivariate and repeated measures analysis. The multivariate analysis revealed a significant difference between groups with pairwise comparisons at the overall .05 level indicating a significant difference between the groups on vestibular domain ($p=.008$). Pairwise comparisons for the vagal responses on tactile domain of the SCP approached significance ($p=.077$). See figure 1. A Mann Whitney U test was performed on change scores to look at between group differences on the recovery domain while controlling for group differences in baseline measures. There was not a significant difference between groups on this domain, Mann Whitney $U = 50.0$, $p=.491$.

Figure 1. Between group differences in vagal responses on the SCP.



Within group differences in vagal tone were analyzed using a repeated measures ANOVA. Maulchy's test found that an assumption of sphericity is plausible (p=.055). The overall test for differences in means was significant, F(5)=3.04, p=.013 with a significant interaction effect between SCP domains and group. Newman Keuls post hoc test revealed a significant difference between groups for the tactile and vestibular domains (r=o). See figure 2.

Figure 2. Within group vagal response patterns on the SCP.



Between group differences on the Short Sensory Profile

A multivariate analysis of variance was conducted to examine between group differences on the SSP. Pairwise comparisons using the Bonferroni method indicate that the groups differ statistically on all sections of the SSP except movement sensitivity. See Table 2.

Table 2. Group differences on the SSP.

SSP	TYP		ASD		Sig. (a)
	M	SD	M	SD	
SSP total	175.0	15.4	128.0	16.4	.000
Tactile	32.3	2.5	28.4	5.1	.013
Taste/Smell	18.5	5.5	11.6	6.2	.011
Movement	13.8	2.4	14.6	.81	.298
Under resp./ seeks sensation	32.3	3.0	18.3	4.2	.000
Auditory Filter	26.9	2.9	17.0	5.7	.000
Low energy	28.6	4.1	21.6	7.7	.012
Visual Auditory	24.2	1.7	16.4	4.5	.000

a Adjustment for multiple comparisons: Bonferroni

significant at the .05 level.

Correlations between vagal measures and SSP scores

Pearson's correlation coefficient was used to examine relationships between vagal tone measures on the SCP and sections of the SSP. When conducting the analysis with groups and sessions combined we find the vestibular domain is correlated to total (r=.510) and auditory (r=.526) sections of the SSP. We also found that the visual auditory section was correlated with the vagal scores on the olfaction domain (r=.509), tactile domain (r=.558), vestibular domain (r=.668) and average response (r=.542). When conducting the on each group separately but with session spooled, there were no relationships at r = ≥ .5 found for the typically developing group. Only one linear relationship was noted for the ASD group and that was between the visual/auditory section of the SSP and the vestibular domain of the SCP with an r=.569. Interestingly, when each session was analyzed separately for the ASD group, the visual/auditory section of the SSP was found to be correlated with the following SCP domains in session 2: tones (r=.627), sirens (r=.555), olfaction (r=.606), tactile (r=.551), vestibular (r=.708) and average response (r=.613). The typically developing group did not demonstrate any relationships. Correlations of the SSP to recovery domain for the ASD group were performed using a Spearman Rho correlation coefficient. There were no significant correlations. See Table 3 for significant correlations.

Table 3. Correlations between SSP sections and vagal responses on domains of SCP.

SSP	SCP domain	r
TYP/ASD sessions combined		
SSP total	vestibular	.510
Auditory	vestibular	.526
Visual/auditory	vestibular	.668
Visual/auditory	olfaction	.509
Visual/auditory	tactile	.558
Visual/auditory	av. response	.542
ASD sessions combined		
Visual/auditory	vestibular	.569
ASD session 2		
Visual/auditory	tones (2)	.627
Visual/auditory	sirens (2)	.555
Visual/auditory	olfaction (2)	.606
Visual/auditory	tactile (2)	.551
Visual/auditory	vestibular (2)	.708
Visual/auditory	av. response	.613

* Correlation is significant at the .01 level (2tailed)

Additionally, 99.9% of the SCP domains were highly correlated ($r>.75$) across both testing sessions in the group of children with ASD, with the exception being visual 1 to vestibular 2 which yielded an r of .629. For example baseline 2 is highly correlated to tones 1 ($r=.893$). Only 77.5% of the SCP domains across both testing sessions for the typically developing group demonstrated a significant linear relationship with an r greater than .75.

Chapter V

Discussion

The results of this study provide preliminary support for the hypothesis that children with ASD demonstrate less physiologic flexibility than typically developing children. This is supported by several findings from this study. First, the children with ASD demonstrated less of a change in vagal tone across sessions thereby demonstrating greater reliability in responses within a very narrow range (.819-.916) as compared to the typically developing children. Since changes in vagal tone are an index physiologic flexibility this finding suggests that children with ASD do not vary their physiologic responses to environmental events . Second, the children with ASD demonstrated a statistically significant lower baseline vagal tone (less heart rate variability at rest) than did typically developing children. Third, the children with ASD demonstrated statistically significant lower vagal scores in response to vestibular stimulation as administered via the SCP. The children with ASD also demonstrated lower vagal tone in response to ALL the other sensations in the SCP although this was not statistically significant. Further, the group of children with ASD demonstrated very little variability across sessions as evidenced by the fact that 99.9% of the SCP domains across sessions were highly correlated to each other.

According to the theory of neuro-occupation, the autonomic nervous system (ANS) is thought to provide the foundation for behavioral regulation through physiological responses to both internal and environmental demands. This regulation supports our ability to engage in activities and interactions and is thus foundational to occupational performance. Based on the results reported here it is reasonable to suggest that decreased physiologic flexibility may be the cause of the decreased behavioral flexibility, characteristic of children with ASD.

This study also provides preliminary support for the relationship between physiology and behavior. First, between group differences on the domains of the SCP demonstrate that typically developing children and children with ASD respond differently (statistically) to vestibular information. The children with autism demonstrated less reactivity to vestibular information. This result, when paired with the finding that the children with autism had higher scores on the movement sensitivity section of the SSP, indicates that these children were less reactive to vestibular information. This supports a relationship between physiologic responsivity to sensation and behavioral responsivity to sensation. In addition, there were several correlations found between vagal responses on the domains of the SCP and sections of the SSP, lending additional support to the possibility of this relationship.

Reliability

As a group, the typically developing children demonstrated more variability that is less consistency, in their vagal responses across the domains of the SCP, while the group of children with ASD had less variability under the same conditions. According to the theory of neuro-occupation the autonomic nervous system is thought to support our ability to engage in activities and interactions. Given the greater variability in vagal tone seen in the typically developing children it is reasonable to hypothesize that this group has greater heart rate variability and thus greater physiologic flexibility. Taken further, it is also reasonable to suggest that this degree of physiologic flexibility better equips these children to handle a variety of environmental challenges and yield adaptive responses. However, it must be noted that the baseline, recovery and average response scores for this group were reliable indicating stability in their basal vagal tone and their responses overall, demonstrating the ability to make quick minute to minute changes in responsivity that are needed for adaptation to the environment. On the other hand, the group of children with autism demonstrated less variability (i.e. greater statistical reliability) than the control group from session 1 to session 2 indicating less heart rate variability between sessions. This decreased physiologic flexibility may be at the root of the decreased behavioral flexibility, characteristic of children with ASD.

Between group differences in vagal responses

The literature shows that children with suspected disruptions in sensory processing demonstrate lower baseline vagal tone (Boccia and Roberts, 2000; Schaaf et al.,2003; Schaaf et al., 2010). The results of this study further support these findings. There was a statistically significant between group difference in baseline vagal tone scores with the group of children with ASD having lower baseline measures. In addition, between group differences on the domains of the SCP demonstrate that typically developing children and children with ASD respond differently (statistically) to vestibular information (p=.013) and to tactile information (p=.077) although the latter difference was not statistically significant. This finding, while exciting is not surprising. Given that stereotyped behaviors characteristic of children with ASD are thought to be sensory driven, specifically body sense (vestibular, proprioceptive and tactile) driven, the statistical difference between groups on the vestibular domain is not unexpected. The vestibular sense, of all the of the body senses, plays a critical role in providing a reference base against which all sensory input and motor output must be evaluated (Cool, 1987). The vestibular system also plays a unique role in regulatory sensory processing in that it is involved in sensory processing and motor output, both key components of RSP. Dr. Ayres (1979) suggested that the vestibular system is the "bridge" between sensory inputs and motor outputs. As discussed in this paper, children with ASD demonstrate behaviors consistent with RSPD (Adrien, et. al, 1987;

Eaves, Ho, & Eaves, 1994; Kientz & Dunn, 1996; Willemsen-Swinkles et al, 1998; Baraneck ,1999; Bagnato & Neilsworth, 1999). Many of the behaviors of children with autism suggest that they are seeking out vestibular and proprioceptive stimulation (Bender, 1947, 1956). For example, children may roll their heads to get vestibular input or flap their hands for proprioceptive input. Freeman et al. (1976) studied the effects of response contingent vestibular stimulation on children with autism versus children with mental retardation. The results demonstrated that children with autism sought out the vestibular stimulation at faster frequencies than children with mental retardation indicating inefficient processing of vestibular information. Miller et al. (2001) found that children with autism spectrum disorders demonstrated hypo-responsivity to sensation as evidenced by a decreased magnitude of EDR's as compared to typically developing children. The results of this study support these findings. The group of children with autism demonstrated less physiologic reactivity to the vestibular information as compared to the typically developing children. See figure 1. Kientz and Dunn (1996) compared children with and without autism on the Sensory Profile and found the behaviors most often seen in children with autism were the same items least frequently seen in children without autism. Our findings support this in that there were statistically significant differences on all sections of the SSP except for movement sensitivity. Based on the available data it appears that the stereotyped behaviors characteristic of children with ASD are not seen in typically developing children. In fact, it is often these stereotyped behaviors that distinguish children with autism from children without autism (Adrien et al., 1987; Kientz & Dunn, 1996). It is possible that some of the stereotyped behaviors seen in children with autism such as body rocking, darting, and head banging are carried out to fulfill a need for additional vestibular input in order to make sense of their environment. Remember, the vestibular system is thought to provide a reference point for all other types of sensory information and is crucial to understanding and relating to one's environment and overall adaptation in one's environment. Inefficient vestibular processing may also contribute to the gross motor and sensorimotor delays that have been reported in children with autism (Baraneck, 1999; Adrien et al.,1987). Lastly, the vestibular system has a direct effect on arousal and muscle tone and as such may account for the statistically significant lower score on the low energy section on the SSP (p=.000) which reflect the effect of arousal and muscle tone on functional behaviors as compared to the typically developing children. This result supports Miller et. al, (2000) findings.

Hence, it can be speculated that typically developing children demonstrate appropriate levels of vagal responsivity to vestibular information and thus do not need to seek it out. Conversely, children with ASD do not adequately respond to the vestibular information and therefore need to seek it out which results in the stereotyped behaviors seen in this population.

Additionally, we saw that as a group, the children with ASD demonstrated less variability that is more consistency, in their vagal scores across domains of the SCP, while the group of typically developing children had more variability under the same conditions (figure1). It is also interesting to note that with both the raw scores and the scores normalized to control for differences in baseline measures, children with ASD demonstrate lower vagal scores on ALL domains of the SCP regardless of significance. With a greater sample we may see these trends reach significance. These findings further support the notion that children with autistic spectrum disorders may have decreased physiologic flexibility which may result in the decreased behavioral flexibility characteristic of regulatory sensory processing disorders and stereotyped behaviors, seen in this population.

Between group differences on SSP

As is seen in Table 2 there were significant differences on all sections of the SSP between the two groups, with the ASD group scoring lower on every section, except *Movement Sensitivity*. The reason that the typically developing group may have scored lower on this section is that these questions relate to general safety awareness. It may be that typically developing children are more "sensitive" to movement because they are more safety aware and have better body awareness than children with ASD who, by report, lack this safety awareness and body awareness that accompanies efficient vestibular processing. The difference in vagal responsivity between the groups support this finding. The typically developing children are more physiologically reactive to vestibular information as compared to the group of the children with ASD. It is this sensitivity or reactivity that distinguishes the groups and may be at the root of the stereotyped behaviors identified in the diagnostic criteria for ASD. Take for example the stereotyped behavior of body rocking. Clinically, it assumed that body rocking is the child's attempt to seek out vestibular input as a result of a hypo responsiveness to vestibular information. The results of this study provide preliminary evidence to support this clinical assumption about the relationship between physiology and behavior.

Correlations between vagal responses and SSP scores

When the data is viewed with groups and sessions combined we find the vestibular domain is correlated to the total and auditory sections of the SSP. Speculation can be made that since the vestibular system is thought to play a critical role in the processing of all sensory information that a relationship between it and the total score of the short sensory profile would exist. We also know that the vestibular system is related to the auditory system in many ways and in fact they share a cranial nerve. So a relationship here is not surprising either. An interesting finding of this study however, is the correlations between the visual auditory section of the SSP and the vagal responses on the domains of the SCP. When data is viewed with session and group combined we find the visual auditory section of the SSP correlated to the vagal tone scores on the olfaction domain, tactile domain, vestibular domain and average response. That is, regardless of group or session, these relationships exist. When the data is viewed with the groups separated, but sessions collapsed, the children with ASD demonstrate a linear relationship between the visual auditory section of the SSP and the vagal responses on the vestibular domain of the SCP. When we looked at session 1 and 2 *separately* we found correlations between the visual auditory section to all of the domains of the SCP except for baseline and visual in the group of children with ASD. A plausible reason for the differences between sessions is that while the responses from session 1 to session 2 were not statistically different, they were just different enough to reveal these correlations. We can speculate that the correlations noted in session 2 may be due to the children's familiarity with the testing protocol. The vagal tone scores from session 1 may reflect the children's response to the novelty of the first testing session. Therefore the vagal scores for the ASD group from session 2 may have been more of a true measure of this group's physiologic responsivity. The visual/auditory section of the SSP contains items that predominately measure responses that are single sensory system based. As opposed to items in other sections that are resultant of multimodal sensory processing. Thus, the items in the visual auditory section of the SSP may have more of a relationship with the domains of the SCP that strive to be single sensory based. To our knowledge, this is the first study that has documented a relationship between physiologic responsivity to sensation, as administered through the SCP, and behavioral responsivity to sensation as measured via the SSP. Replication of this study with a larger sample size is needed to strengthen these findings.

Lastly, the fact that 99.9% of the SCP domains were highly correlated (r>.75) across both testing sessions in the group of children with ASD as compared to 77.5% of the typically developing group supports the fact that children with ASD demonstrate less physiologic variability across time . This in turn supports the hypothesis that decreased physiologic flexibility is associated with the decreased behavioral flexibility characteristic of this population.

Limitations

There are several limitations to this study that may have affected the results. First, the sample size is small and thus findings need to be replicated with a larger sample size. A total of thirty nine boys with and without autism were tested (n=16 typically developing boys and 23 boys with ASD). However, several data files could not be analyzed due to excessive artifact (n= 4 subjects with autism) or technical difficulties (n=2 subjects with ASD and n= 1 control). Further, several testing sessions had to be terminated due to inability of the subject to tolerate the protocol (n=6 subjects with autism). This left data from twenty six boys, eleven with autism and fifteen typically developing. Second, this was a sample of convenience where children were primarily recruited from schools and therapy clinics where the research assistants worked. Given this it was not possible to exactly age match the groups resulting in a significant age difference between the groups t(24) =2.43, p=.023, which is another limitation. Another consideration is the possibility of a learning effect between the two sessions which may have affected the results. Therefore devising another protocol that mimics the sensations administered in the SCP but in a different fashion may be warranted. Another factor that may have affected the results is the possibility that the children experienced anxiety during the first testing session due to the novelty of the situation. Although there was not a significant difference in vagal scores between sessions, within each group, as was seen in the correlations, when each session was viewed separately, there may have been just enough of a difference to affect results. This is consistent with the findings of Schaaf et al. (2010). Thus, it may be prudent to attempt to control for this initial testing anxiety in future studies using this protocol. Lastly, the SCP administers sensation in a controlled manner which lends itself to being used in research protocols. However, the sensations that are being administered are primarily single sensory system based so that comparing results of the physiologic data collected during these experiences to behavioral measures of sensory processing ,which by nature are multisensory, is difficult. The addition of some multisensory experiences to the SCP may increase the ability to more clearly detect correlations between physiologic and behavioral measures of sensory processing. Our findings suggest that there are relationships between the items on the SSP that are primarily single system based to the vagal responses to the sensations of the SCP which are also primarily single system based. Therefore, it is reasonable to postulate that the sensory experiences that are multimodal based may demonstrate relationships to behavioral responses to sensation that are also multimodal.

Chapter VI

Conclusion

To date there have been no studies published that have investigated the reliability of vagal tone in response to sensation across time in typically developing children or children with autistic spectrum disorders. This study gives preliminary information on the stability of vagal responses over time to sensory information in both typically developing children and children with autistic spectrum disorders. Highlighting the fact that children with autistic spectrum disorder demonstrate greater statistical reliability i.e.: less variability in physiologic responsivity from session 1 to session 2, than typically developing children. This study also adds to the information on the physiologic differences in responsivity. The group of children with ASD demonstrate significantly lower baseline vagal tone as compared to the typically developing children which support previous findings of baseline vagal tone measures in groups of children with proposed RSPD (Schaaf et al., 2003; 2010). In addition, we found that children with ASD demonstrate less physiologic reactivity to vestibular information than do the typically developing children. This decreased reactivity to vestibular information is also seen behaviorally in the scores on the movement sensitivity section of the SSP and hence point to a relationship between physiology and behavior. This relationship is also supported by the correlations found between vagal responses to sensation administered via the SCP and behavioral responses to sensation as measured via the SSP. Combined these findings provides preliminary evidence to support the theoretical and clinical assumptions that neurophysiologic state affects behavior.

Based on the theory of neuro-occupation, this study provides insights into the possible physiologic causes of the regulatory sensory processing disorders and behavioral rigidity seen in children with autistic spectrum disorder. Future research is needed to strengthen the results of this study using a larger sample size. There are many directions that this line of research can take however one exciting direction we can now follow based on the preliminary information we have on the stability of vagal tone responses to sensation over time, is to conduct intervention studies to determine whether certain targeted interventions can change autonomic responsivity and thus behavior.

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Appendix A

RSPD Classification System

Type	*Behavioral Characteristics*
Sensory Modulation Challenges – Type I	
Over-responsive, fearful, anxious	Difficulty with transitions Poor adaptability Poor socialization
Over-responsive, negative, stubborn	Controlling Aggressive, impulsive Defiant
Under-responsive, self-absorbed	Quiet, passive or withdrawn May appear disinterested Easily exhausted
Self-absorbed, creative	Highly intelligent and creative Escapes in fantasy when faced with challenges Easily distracted/ appears inattentive
Active, sensory seeking	Constantly in motion Impulsive May insist on getting own way
Sensory Discrimination Challenges – Type II	
Sensory discrimination challenges	Specific system challenges – visual, auditory, tactile, olfactory, proprioceptive, vestibular
Sensory Based Motor Challenges – Type III	
Postural Challenges	Poor trunk stability Either avoidance of movement or very physically active but lacking control and safety awareness Visual distractibility due to problems maintaining visual focus
Dyspraxia	Poor in sports and participating on a team Takes more time and practice to learn a new skill Difficulty with fine motor manipulative activities such as scissor use and writing with a pencil
Mixed Regulatory Sensory Processing Patterns	
Sensory over-responsivity with sensory seeking	
Sensory over-responsivity with sudden under-responsivity	
Sensory over-responsivity with sensory discrimination difficulties	
Sensory over-responsivity with dyspraxia	
Sensory under-responsivity with sensory over-responsivity	
Sensory under-responsivity with dyspraxia	
Sensory discrimination problems with dyspraxia	

Appendix B

Letter of Approval from IRB

Dear Ms Caveliere,

SETON HALL UNIVERSITY

April 11, 2006

Catherine Cavelier
8 Valley Place
Tenafly NJ 07670

Thank you for you cooperation

The Seton Hall University Institutional Review Board has reviewed and approved as submitted under expedited review your research proposal entitled "Reliability of Physiologic Response to Sensation in Children with and without Autism". The IRB reserves the right to recall the proposal at any time for full review.

Enclosed for your records are the signed Request for Approval form and the stamped original Consent Form, Assent Form and recruitment flyer. Make copies only of these stamped forms.

The Institutional Review Board approval of your research is valid for a one-year period from the date of this letter. During this time, any changes to the research protocol must be reviewed and approved by the IRB prior to their implementation.

According to federal regulations, continuing review of already approved research is mandated to take place at least 12 months after this initial approval. You will receive communication from the IRB Office for this several months before the anniversary date of your initial approval.

Sincerely,

Mary F. Ruzicka, Ph.D

Mary F. Ruzicka, Ph.D
Professor
Director, Institutional Review Board

Susan Simpkins, EdD, PT

Office of Institutional Review Board
Presidents Hall
Tel. 973.313.6314 • Fax 973.275.2361
400 South Orange Avenue • South Orange, New Jersey 07079-2641

Appendix C
Letter of Solicitation

SETON HALL UNIVERSITY
1 8 5 6

Dear Parents/Guardians,

My name is Catherine Cavaliere. I am an Occupational Therapist in the doctoral program in Health Sciences at Seton Hall University and am conducting a research project in partial fulfillment of the requirement for my PhD titled *Reliability of Physiologic Responses to Sensation in Children with and without Autistic Spectrum Disorders.*

The purpose of the study is to investigate how children's bodies respond to sensation. It is important to study children's responses to sensation in order for us to better understand exactly how children process sensory information and to provide a foundation upon which we can develop and test treatment strategies aimed at reducing abnormal responses to sensation.

As you know, an overwhelming number of children both with and without autistic spectrum disorders experience sensory processing difficulties. These include over responsiveness and under responsiveness to sensation including auditory information, tactile (touch) information and movement. Problems with sensory processing can affect social and communication skills and have a direct impact on learning.

This study will evaluate children's responses to touch, smell, sight, sound and movement. We will measure heart rate and sweat gland activity while your child is exposed to various sensory experiences similar to the sensory experiences that your child is exposed to everyday. If your child indicates that he wants to stop or if you would like the testing to be stopped, the testing will be halted *immediately.* Participation will require two sessions approximately 45 minutes each.

For your convenience we will be conducting the testing at Celebrate the Children. You the parent are welcome to be present for the testing. We will notify you in advance of the day and time that your child is scheduled to participate.

We are looking for boys with autism spectrum disorders 4-11 years. We are also looking for typically developing boys within the same age range. To be eligible for the study the child must, be free of any other medical or psychological disorders, not currently taking medication that affects arousal level, be able to follow simple directions and be able to remain seated for a 30 minute period with occasional breaks.

Your child will receive a small gift at the end of each testing session for his participation and each family will receive a gift certificate to a local chain restaurant after the final testing session.

All information about your child will remain *confidential.* Participation in this study is completely *voluntary.* You may refuse to have your child participate or withdraw permission at any time.

By participating in this study you and your child will be making a significant contribution to the effort to better understanding autism. .

If you would like additional information on this study or you would like your child to participate in the study please fill out and return the below portion of this letter to your child's therapist.

You may also contact me directly by email at cmcavaliere@optonline.net or by phone at 551-587-1249.

Thank you for your time and consideration.

Catherine Cavaliere, OTR/L

__________ Yes, I would like additional information about this study / am interested in having my child participate in this study

Parent's name __

Child's name __

Phone number __

Email address _

Appendix D

Flyer

Children Needed for a Study on Sensory Processing

We are investigating how children with and without autism spectrum disorders respond to sensory information.

This study will evaluate children's responses to touch, smell, sight, sound and movement. We will measure heart rate and sweat gland activity while your child is exposed to various sensory experiences similar to the sensory experiences that your child is exposed to everyday.

We will arrange to test your child at a location and time that is convenient for you. Participation will require two visits of approximately one hour each.

Your child will receive a small gift at the end of each testing session for his participation.

Eligibility Requirements:

- Typically developing boys AND boys with autistic spectrum disorders
- Ages 4-11 years
- Free of any medical or psychological disorders
- Not currently taking any medications known to affect arousal level
- Able to follow simple directions
- Able to remain seated for approximately 30 minutes with breaks as needed

Participation in this study is completely voluntary. Care will be given to preserve the confidentiality of all information that you give us. All forms and videotapes with your child's name on it will be kept in a locked file cabinet at Seton Hall University. Only the researchers will have access to these files and videotapes. Each child will receive a special code for data collection purposes to assure anonymity. All information will be kept for three years and then shredded.

FOR MORE INFORMATION ON THIS STUDY OR TO SCHEDULE A TIME FOR TESTING PLEASE CONTACT:

Cathy Cavaliere, OTR/L
Seton Hall University
551-587-1249
cmcavaliere@optonline.net

Appendix E
Identifying Information

Identifying Information

Study Title: Reliability of Physiologic Responses to Sensation in Children with and without Autistic Spectrum Disorders

Child's name

Parent/s name/s

Address

Phone number

Child's age

If your child has autism, when did he receive the diagnosis?

Does your child have any other medical diagnosis?

Does your child have a seizure disorder or has your child ever experienced seizures? Please explain

Is your child currently taking any medications? If so please list

Has your child had a hearing test in the recent past? What was the result of that test?

School name/location

Is your child in a self-contained classroom (only special education students) or in an inclusive classroom (both regular education and special education students)

Does your child currently receive any therapies or has received any therapies in the past (ie: OT, PT, Speech, ABA)? If so please specify ______

Has your child ever received any alternative therapies such as Auditory Integration Training or Nutritional Therapy? If so please specify ______

Does your child have any apparent sensitivities to sensation? If so please specify ______

Does your child understand simple commands such as "Bring me the book"? ______

Does your child verbally or gesturally respond to your questions such as "Would you like milk or juice?"? ______

What are your child's favorite things to do/hobbies? Please be specific. For example: my child love to watch blues clues

Child's Doctor's name/sand phone number ______

Other pertinent information______

Appendix F
Consent

SETON HALL UNIVERSITY
1 8 5 6

Consent Form

Study Title: Reliability of Physiologic Responses to Sensation in Children with and without Autism
The primary investigator for this research is Catherine Cavaliere, OTR/L. Catherine is an Occupational Therapist who is in the Doctoral Program in Health Sciences at Seton Hall University.

Purpose:
The purpose of this study is to evaluate the reliability of physiologic measures of sensory processing in children. We will record children's heart rate and sweat gland responses to touch, smell, sight, sound and movement, similar to the sensory experiences that your child is exposed to everyday. It is important to study children's responses to sensation in order for us to better understand exactly how children process sensory information and to provide a foundation upon which we can develop and test treatment strategies aimed at reducing abnormal responses in children that may help these children function better at home, school and in the community. The total procedure time is approximately one hour. Forty children will participate in the study. Twenty of the children will have autism and twenty of the children will not have autism.

Procedures:
You, the parent/guardian, and your child will come to either the Human Performance Lab at Seton Hall University, Barpak Health Services in Bergenfield, New Jersey or the Teaneck Community Charter School in Teaneck, NJ. You and your child will be asked to come two times. You will be asked to fill out the Short Sensory Profile, a 46-item caregiver questionnaire about your child's behavioral responses to sensation such as your child's ability to pay attention or eat a variety of foods, at your initial visit. This part should take approximately 20 minutes. While you are filling out the Short Sensory Profile, the researcher will be acquainting your child to the laboratory setting, which is designed to look like a spaceship. Once your child is comfortable in the lab we will measure your child's heart rate and sweat gland activity while we have him smell

wintergreen oil, touch his chin lightly with a feather, look at a series of blinking lights, listen to an audio tape of siren sounds, and tip him slowly back in a chair. We will measure heart rate by placing three electrodes on your child's chest.

We will measure sweat gland activity by placing two small electrodes on your child's hand. You may remain in the room with your child the entire time. If your child indicates that he wants to stop the testing will be halted immediately. Each session will take approximately 30 minutes. The total time for the first session including introducing your child to the spaceship room, completion of the Short Sensory Profile and the administration of the sensory stimulation should take approximately one hour. All subsequent sessions should take approximately 45 minutes. Each session will be videotaped.

Confidentiality:
Care will be given to preserve the confidentiality of all information that you give us. All forms and videotapes with your child's name on it will be kept in a locked file cabinet at Seton Hall University. Only the researchers will have access to these files and videotapes.

Anonymity:
Each child will receive a special code for data collection purposes to assure anonymity. All information will be kept for three years and then shredded.

Potential Risks:
None of the procedures are painful, although it is possible that some children may find some sensations uncomfortable. If this occurs we will immediately halt the testing.

Potential Benefits:
Your child will not benefit directly from participation in this study. However, the findings of this study may help define how children respond to sensation as well as lay a foundation for future studies in the treatment of sensory processing disorders.

Alternative treatments:
The alternative to your child participating in this study is to not participate in this study. This is not a treatment study and therefore does not affect any current treatment procedures that your child may be involved in.

Videotape:
Each testing session will be videotaped. You the parent/guardian and your child have the right to request to view the tape and may request the tape be destroyed at any time. All videotapes will be coded to assure anonymity. All videotapes will be destroyed at the end of a three-year period.

Voluntary nature of study:
Participation in this study is completely voluntary. You or your child may refuse participation or withdraw permission at any time. If your child does participate, you or your child may request that the testing be stopped at any time.

You will be given a copy of this signed and dated informed consent form stating that you give permission for your child to participate in the study.

If you have any questions or concerns about this study please contact the principal investigator, Catherine Cavaliere, by email at cmcavaliere@optonline.net or by phone at551-587-1249. If you have any additional questions or concerns you may contact Dr. Susan Simpkins, Research Advisor at 973-275-2916 or the Institutional Review Board at 973-313-6314.

Parent/Legal Guardian's name Child's name

Parent/Legal Guradian's signature Date

Subject or Authorized Representative Date

School of Graduate Medical Education
Department of Graduate Programs in Health Sciences
Tel 973 275 2076 Fax 973 275 2370 TDD 973 275 2169
400 South Orange Avenue South Orange, New Jersey 07079 ***gradmeded.shu.edu***

Appendix G
Assent

SETON HALL UNIVERSITY
8 5 6

Assent

Study Title: Reliability of Physiologic Responses to Sensation in Children with and without Autism

My name is Cathy Cavaliere. I am an Occupational Therapist. I am a student just like you. I go to school at Seton Hall University. I am in charge of the testing. The reason we are doing the testing is to see how children's bodies react to touching things, smelling things, seeing things, hearing things and moving.

You and your mom/dad/guardian will come to a special room at either the Human Performance Lab at Seton Hall University, Barpak Health Services in Bergenfield, New Jersey or the Teaneck Community Charter School in Teaneck, NJ.. This room looks like a pretend spaceship! You will be asked to come two times. Your mom/dad/guardian will be asked questions about you such as your ability to pay attention or the kinds of foods you like to eat. When you are ready we will look at your heart beat and how you sweat while we have you smell something, touch your chin lightly with a feather, look at blinking lights, listen to a tape of siren sounds, and tip you slowly back in a chair, like a rocking chair. We will look at your heart beat by placing three stickers with wires on them on your chest and how much you sweat by placing two tiny stickers on your hand (show child electrodes). The stickers do not hurt. (Possibly let child try one on) Your mom/dad/guardian can be in the room with you the whole time. If you want to stop the testing let us know and we will stop right away. You will be in the spaceship room for about one hour each time you come. You will be videotaped each time that you come to the spaceship room.

When you are ready we will look at your heart beat and how you sweat while we have you smell something, touch your chin lightly with a feather, look at blinking lights, listen to a tape of siren sounds, and tip you slowly back in a chair, like a rocking chair. We will look at your heart beat by placing three stickers with wires on them on your chest and how much you sweat by placing two tiny stickers on your hand (show child electrodes). The stickers do not hurt. (Possibly let child try one on) Your mom/dad/guardian can be in the room with you the whole time. If you want to stop the testing let us know and we will stop right away. You will be in the spaceship room for about one hour each time you come. You will be videotaped each time that you come to the spaceship room.

Any information that you or your mom/dad/guardian gives us will be kept in a locked file cabinet at Seton Hall University. No one will see this information except for me and the people I work with in the spaceship room.

You will receive a special code for the heart beat and sweat information that we take from you so that other people will not know that it is yours. All information will be kept for three years and then we will throw it away.

Nothing we do today will hurt you, although it is possible that you will not like some things that you see, hear, smell or feel. If this happens let us know right away and we will stop the testing.

You will not receive anything special for coming to the spaceship room but by coming you may help us find ways to help children who have problems with the certain things they see, hear, feel or smell.

You do not have to come to the spaceship room if you do not want to. No one will be mad at you if you choose not to.

Each time you come here you will be videotaped. You or your mom/dad/guardian has the right to request to see the tape and may ask that the tape be thrown away at any time. The videotape will also receive a special code so that we do not know it is yours.
All videotapes will be thrown away at the end of a three-year period.

You have the right to say that you do not want to come to the spaceship room. You also have the right to change your mind and stop coming to the spaceship room at any time. If you do come to the spaceship room, you have the right to stop testing at any time.

You and your mom/dad/guardian will be given a copy of this paper stating that you want to come to the spaceship room and see, hear and feel some things.

If you have any questions about this study you may contact me, Catherine Cavaliere, by email at cmcavaliere@optonline.net or by phone at 551-587-1249. If you have any further questions or concerns about this study you may contact Dr. Susan Simpkins, Research Advisor at 973-275-2916 or the Institutional Review Board at 973-313-6314.

Child's name

Parent/Legal Guardian's name | Child's signature

Parent/Legal Guardian's signature | Date

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Department of Graduate Programs in Health Sciences
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