Caregiver Burden, Participation, and Sensory Subtypes in Children with Autism

DISSERTATION

Presented in Partial Fulfillment of the Requirements for the Degree Doctor of Philosophy in the Graduate School of The Ohio State University

By

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Abstract

Fifty-three percent of occupational therapists lack confidence when working with the autism population and 60% consider autism spectrum disorders (ASD) to be more challenging to work with than other diagnoses (Ashburner, Rodger, Ziviani, & Jones, 2014). One possible reason for this is the heterogeneity in clinical presentation in ASD, as interventions may work for some children with ASD but not others (Hoddenbach et al., 2012). To narrow intervention focus and support clinical reasoning, there is a widespread effort to identify homogenous subtypes of children with ASD who may respond similarly to intervention (The Interagency Autism Coordinating Committee, 2014).

One such subtyping schema was proposed by Lane et al. (2014) and consists of four sensory subtypes of children with ASD based on parent responses to the 38-item Short Sensory Profile (SSP): 1) Sensory Adaptive; 2) Taste/Smell Sensitive; 3) Postural Inattentive; and 4) Generalized Sensory Difference. Differences between sensory subtypes have been documented in three out of five health-related domains of the International Classification of Functioning, Disability and Health (ICF), including body functions and structures, activities, and personal factors (Lane et al., 2010, 2011, 2014a; Tanner and Lane, unpublished manuscript). This information may be valuable in the development of decision-making tools for clinicians working with children with ASD and their families. However, differences between the subtypes in the two remaining ICF health-related domains of environmental factors, such as caregiver burden, and child participation in valued activities have yet to be explored. Moreover, our pilot data indicate that not all SSP items are relevant to subtype determination, suggesting that reduced respondent burden and increased efficiency via item reduction may be possible.
Thus, prior to widespread clinical use of sensory subtyping, it is necessary to determine differences between subtypes in caregiver burden, determine differences between subtypes in child participation, and improve the efficiency of the sensory subtyping algorithm via item reduction. The objectives of this dissertation are to: 1) determine differences between subtypes in level of burden that caregivers report; 2) determine differences between subtypes in child participation of valued activities; and 3) reduce the number of SSP items needed for sensory subtyping.

Method: To achieve these objectives, a national survey of 378 caregivers of children with ASD aged 5-13 years was conducted. Caregivers completed the SSP as well as valid and reliable measures of caregiver burden and child participation. Caregiver burden was measured by: 1) the SF-12v2, which is a measure of health related quality of life, and 2) the Caregiver Strain Questionnaire, which is a measure of perceived strain. The Participation and Environment Measure - Children and Youth was used to measure child participation in meaningful activities. Canonical correlation analyses were used to achieve objectives 1 and 2, while a multinomial logistic regression with lasso penalization was used to achieve objective 3.

Results: Results from Chapter 2 revealed that caregivers of children with ASD in different sensory subtypes did not differ in perceived mental and physical health related quality of life, but did differ in level of perceived caregiver strain. When compared with caregivers of children in the Generalized Sensory Difference subtype, caregivers of children in the Sensory Adaptive subtype reported the lowest levels of strain, followed by caregivers of children in the Taste/Smell Sensitive subtype. Caregivers of children in the Postural Inattentive and Generalized Sensory Difference subtypes did not significantly
differ in level of strain. Similarly, results from Chapter 3 indicate that children in different sensory subtypes were found to differ in their participation in home, school, and community activities. When compared with children in the Generalized Sensory Difference subtype, children in the Sensory Adaptive subtype had the highest levels of participation, followed by children in the Taste/Smell Sensitive subtype. The Postural Inattentive and Generalized Sensory Difference subtypes did not significantly differ in level of participation. Findings from Chapter 4 identified a subset of 18 SSP items that determine membership to the Taste/Smell Sensitive, Postural Inattentive, or Generalized Sensory Difference subtypes with 94.6% agreement with the existing algorithm based on the 38-item SSP. This subset does not, however, identify members of the Sensory Adaptive subtype.

Conclusion: Findings of this dissertation were combined with existing literature to create a decision-making framework that focuses the scope of intervention for each sensory subtype by linking deficits with evidence-based interventions. This body of work improves the clinical and research utility of sensory subtyping in ASD by: 1) linking subtypes to areas of caregiver burden and child participation to target for intervention, and 2) reducing respondent burden by determining sensory subtypes with fewer SSP items. This research contributes much needed empirical evidence to guide clinical decision-making in intervention selection for children with ASD. Future studies are necessary to further these findings and effectively build upon this evidence by examining the degree to which this decision-making framework improves treatment outcomes for children with ASD. Increasing evidence in support of this framework will aid in its
clinical translation, where it will likely have a direct positive impact on the quality of care provided for children with ASD.
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Finally, I would like to thank Dr. Alison Lane and Dr. Amy Darragh. I am extremely grateful to Dr. Lane for her years of research mentorship and for serving as the content expert for sensory subtypes in children with autism on my committee. Dr. Lane is truly a role model to me in every sense of the word and I would like to express my deepest gratitude for her part in shaping me into a research scientist. I would also like to express my appreciation to Dr. Darragh for serving as my advisor. The path to completing this dissertation has been circuitous, but she has been unwavering in her support and encouragement.

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Publications


**Fields of Study**

Major Field: Health and Rehabilitation Science

Minor Field: Data Analytics and Research Methodology
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Dissertation Structure

In addition to the requirements of the Graduate School of The Ohio State University, this document has been prepared according to standards set forth by the School of Health and Rehabilitation Sciences handbook (9/2013). Dissertations produced within the Health and Rehabilitation Sciences PhD Program “should be comprised of at least three original, journal level manuscripts” (p.31). The structure and format of this document is as follows:

1. Chapter 1 provides an introduction to the content area.

2. Chapters 2, 3, and 4 contain the required three manuscripts. These chapters are stand-alone manuscripts. As such, repetition may be noted among them.

3. Chapter 5 contains summaries, conclusions, and additional commentary that does not fit within the three, primary manuscripts.

4. APA citation style is used for this document.
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Chapter 1: Introduction

1.1 Autism Spectrum Disorder

Autism spectrum disorder (ASD) is a neurodevelopmental condition characterized by deficits in language and social skills as well as atypical patterns of behavior, movement, interests, and reactions to sensory input (American Psychiatric Association, 2013). Autism spectrum disorders are one of the most common forms of developmental disability in the U.S. with an estimated prevalence of 1-2% of all school-aged children, 1 in 42 boys and 1 in 182 girls (Baio, 2012; Bölte, 2014; Christensen et al., 2016). The cause of ASD remains unknown, however most evidence suggests a combination of genetic predisposition and environmental variables underlie the development of ASD (Lichtenstein, Carlström, Råstam, Gillberg, & Anckarsäter, 2010; Ronald & Hoekstra, 2011).

Individuals with ASD frequently struggle with engagement in many daily activities including self-care, social participation, play, and school or work-related tasks (Miller-Kuhaneck & Watling, 2010). Occupational therapy, which uses everyday life activities therapeutically for the “purpose of enhancing or enabling participation in roles, habits, and routines in home, school, workplace, community, and other settings,” is the second most frequently used therapy for children with ASD (AOTA, 2014, pg. S1; McLennan, Huculak, & Sheehan, 2008). Occupational therapists work with children with ASD to improve their level of participation and independence in daily activities through
the use of a variety of evidence-based interventions (Kuhaneck, Madonna, Novak, & Pearson, 2015; Tanner, Hand, O’Toole, & Lane, 2015; Watling & Hauer, 2015; Weaver, 2015). One barrier to the successful treatment of children with ASD, however, is phenotypic heterogeneity with regard to symptom severity, intellectual functioning, social skills, and comorbidities (Bölte, 2014). This heterogeneity leads to difficulties with intervention selection and intervention efficacy, as interventions may work for some children with ASD but not others (Hoddenbach et al., 2012). As a result, there is a widespread effort to identify homogenous subgroups of children with ASD who may respond similarly to intervention (The Interagency Autism Coordinating Committee, 2014). Many different mechanisms for ASD subtype determination have been posited, including: intellectual functioning; language abilities; style of social interaction; and neurobiological features (Beglinger & Smith, 2005; Bölte, 2014; Goh, Dong, Zhang, DiMauro, & Peterson, 2014; Whitehouse, Barry, & Bishop, 2008). Some of these methods are substantiated by promising preliminary findings (i.e., intellectual functioning, social subtypes), whereas others lack validity (i.e., language subtypes) or lack direct clinical applications (i.e., neurobiological features) (Beglinger & Smith, 2005; Goh et al., 2014; Whitehouse et al., 2008).

Recently, researchers have suggested that behaviors associated with difficulties processing and integrating sensory information, termed sensory features, are another basis upon which subtypes in ASD may be defined (Ausderau et al., 2014; Baranek et al., 2006; Lane, Molloy, & Bishop, 2014). Individuals with ASD and sensory features may: demonstrate hyper- or hypo-reactivity to sensory stimuli; have unusual sensory interests; inaccurately perceive sensory stimuli; and/or have difficulties managing
multiple sensory stimuli simultaneously (Schaaf & Lane, 2015a). For example, a child who is hyper-reactive to sound may cover their ears when a dog barks or when the phone rings, while a child with unusual interests in vestibular stimulation may engage in sensory-stimulating activities such as rocking back and forth or spinning in a circle. Sensory features are common in ASD and have been reported in up to 92% of all individuals with an ASD diagnosis (Schaaf & Lane, 2015a). As a result of this high prevalence, sensory features are now included as one manifestation of restricted and repetitive behaviors in the diagnostic criteria of ASD in the DSM-5 (American Psychiatric Association, 2013). A growing body of literature asserts that distinct sensory subtypes of children with ASD can be identified by examining patterns of sensory features. Table 1.1 summarizes the differences between the terms “sensory features” and “sensory subtypes.”
Table 1.1 Definitions of sensory features and sensory subtypes

<table>
<thead>
<tr>
<th>Term</th>
<th>Definition</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sensory Features</td>
<td>• Behaviors associated with difficulties processing and integrating sensory information</td>
</tr>
<tr>
<td></td>
<td>• Includes:</td>
</tr>
<tr>
<td></td>
<td>• Sensory reactivity (hyper- or hypo-reactivity)</td>
</tr>
<tr>
<td></td>
<td>• Unusual sensory interests</td>
</tr>
<tr>
<td></td>
<td>• Altered perceptions of sensory stimuli</td>
</tr>
<tr>
<td></td>
<td>• Difficulties managing multiple sensory stimuli simultaneously</td>
</tr>
<tr>
<td>Sensory Subtypes</td>
<td>• Groups of children with ASD who demonstrate shared patterns of sensory features</td>
</tr>
</tbody>
</table>

Schaaf & Lane, 2015a
1.2 Sensory Subtypes in ASD

The earliest sensory subtypes were those published by Lane and colleagues using caregiver responses to a questionnaire called the Short Sensory Profile (Lane et al., 2010; Lane, Dennis, & Geraghty, 2011; Lane et al., 2014a). The Short Sensory Profile (SSP) measures sensory processing in seven domains; these domains are summarized in Table 1.2. The four sensory subtypes proposed by Lane et al. (2014a) are: 1) Sensory Adaptive (SA); 2) Taste/Smell Sensitive (TSS); 3) Postural Inattentive (PI); and 4) Generalized Sensory Difference (GSD). Table 1.3 summarizes the four subtypes. These four subtypes differ on the severity and focus of sensory features (Figure 1.1; Lane et al., 2014a). The basis for variation in sensory feature severity is best mapped by the degree of difficulty displayed in the SSP domains of under responsive/seeks sensation and auditory filtering. Those in the SA subtype have only mild difficulties, TSS and PI have intermediate, and GSD have the most difficulty in these SSP domains. The two intermediate-severity subtypes, TSS and PI, are further differentiated by their focus. Specifically, the TSS subtype demonstrates extreme scores in the taste/smell sensitivity domain of the SSP, while the PI subtype demonstrates extreme scores in the low energy/weak domain. Due to the mild nature of their sensory features, it is unlikely that the functional performance of children in the SA subtype is strongly impacted by sensory features. In contrast, the GSD subtype demonstrates clinically meaningful difficulties in all seven sensory domains measured by the SSP, which may have widespread implications for functional performance.
Table 1.2 Descriptions of Short Sensory Profile domains

<table>
<thead>
<tr>
<th>Domain</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Tactile Sensitivity</td>
<td>Measures responses to stimuli that touch the skin</td>
</tr>
<tr>
<td>Taste/Smell Sensitivity</td>
<td>Measures responses to foods, food textures, food temperatures, and food smells</td>
</tr>
<tr>
<td>Movement Sensitivity</td>
<td>Measures responses to movement and movement-related activities</td>
</tr>
<tr>
<td>Under-responsive/Seeks Sensation</td>
<td>Measures decreased responsiveness to sensory stimuli as well as behaviors associated with seeking out additional sensory stimulation</td>
</tr>
<tr>
<td>Auditory Filtering</td>
<td>Measures a child’s ability to concentrate with background noise and attend to salient auditory stimuli</td>
</tr>
<tr>
<td>Low Energy/Weak</td>
<td>Measures a child’s strength, endurance, and postural control</td>
</tr>
<tr>
<td>Visual/Auditory Sensitivity</td>
<td>Measures responses to visual stimuli, such as bright lights, and auditory stimuli such as a dog barking or vacuum cleaner</td>
</tr>
</tbody>
</table>
Table 1.3 Descriptions of sensory subtypes

<table>
<thead>
<tr>
<th>Subtype</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sensory Adaptive</td>
<td>• Typical function in five of the seven sensory domains</td>
</tr>
<tr>
<td></td>
<td>• Mild difficulties in auditory filtering and under-responsive/seeks sensation</td>
</tr>
<tr>
<td></td>
<td>• Unlikely that functional performance is strongly impacted by sensory differences</td>
</tr>
<tr>
<td>Taste/Smell Sensitive</td>
<td>• Taste/smell sensitivity</td>
</tr>
<tr>
<td></td>
<td>• Clinically meaningful concerns in auditory filtering and under-responsive/seeks sensation</td>
</tr>
<tr>
<td>Postural Inattentive</td>
<td>• Extreme score in low energy/weak domain</td>
</tr>
<tr>
<td></td>
<td>• Clinically meaningful concerns in auditory filtering and under-responsive/seeks sensation</td>
</tr>
<tr>
<td>Generalized Sensory Difference</td>
<td>• Affected in all sensory domains</td>
</tr>
<tr>
<td></td>
<td>• Only subtype to have clinically meaningful concerns in movement sensitivity</td>
</tr>
</tbody>
</table>
Figure 1.1 Relationships of sensory subtypes to severity and focus of sensory features

Note. SA = Sensory Adaptive; TSS = Taste/Smell Sensitive; PI = Postural Inattentive; GSD = Generalized Sensory Difference
Other research groups have proposed alternate sensory subtyping models. Ausderau and colleagues (2014) also proposed a four-subtype model using a parent report measure: 1) Mild; 2) Sensitive-Distressed; 3) Attenuated-Preoccupied; and 4) Extreme-Mixed. In this model, four types of sensory features, extracted from the Sensory Experiences Questionnaire (SEQ), were used to characterize sensory subtypes (Baranek, 2009). The sensory features used in this model were: 1) sensory interests, repetitions, and seeking behaviors; 2) hypo-reactivity; 3) hyper-reactivity; and 4) enhanced sensory perception. The Mild subtype is characterized by low scores on each of the four sensory features. Children in the Sensitive-Distressed subtype demonstrate hyper-reactivity and enhanced sensory perception, while the Attenuated-Preoccupied subtype is characterized by hypo-reactivity and sensory interests, repetitions, and seeking behaviors. The Extreme-Mixed subtype displays difficulty with all four sensory patterns.

Despite being derived from different tools, there is a key similarity between this subtyping model and that proposed by Lane and colleagues; both models consist of four subtypes in which one subtype has mild sensory difficulties and another has extreme sensory difficulties. The congruence between these two subtyping methodologies lends support to the validity of sensory subtypes in ASD based on the severity of sensory features. These two classification schemas differ, however, in that Ausderau et al.’s model does not take sensory modality (i.e., tactile, auditory, taste, etc.) into account. In contrast, Lane et al.’s subtypes identify groups of children who demonstrate shared patterns of sensory features in the same sensory domains. For example, children in the TSS subtype demonstrate clinically meaningful hyper-reactivity only to tastes and smells,
whereas children in the GSD subtype demonstrate hyper-reactivity to movement, visual, auditory, tactile, taste, and smell stimuli.

The use of sensory subtyping has the potential to refine diagnosis and intervention selection processes for children with ASD. Identifying subtypes of children with ASD with characteristic patterns of sensory features and associating those patterns with functional performance limitations can guide clinical decision-making and intervention selection, and may subsequently improve intervention effectiveness. As Lane et al.’s sensory subtypes are the most extensively studied, have been replicated in three independent samples, and are widely documented to display differences in various aspects of daily functioning, this sensory subtyping schema was selected for use in the present dissertation (Lane et al., 2010, 2011, 2014a; Tanner & Lane, unpublished manuscript). For the remainder of this document, the phrase “sensory subtypes” will be used to refer to the subtypes proposed by Lane and colleagues, unless otherwise noted.

1.3 Known Differences Between the Sensory Subtypes

Preliminary work indicates that there are distinct non-sensory behavior profiles associated with each of the sensory subtypes. For example, children in the TSS subtype have more severe communication impairments and are picker eaters, while children in the GSD subtype demonstrate more maladaptive behaviors (Lane et al., 2010, 2011). Additionally, differences have been found between the subtypes on daily living skills, emotional regulation and attention (Tanner & Lane, unpublished manuscript). Despite differences in clinical presentation, the four subtypes are not well explained by systematic variation in age, non-verbal IQ, or severity of autistic symptoms such as social interaction, communication, restricted and repetitive behaviors (Lane et al., 2014a).
To better understand known differences between the subtypes, it may be useful to examine existing literature in the context of the International Classification of Functioning, Disability, and Health (ICF). The ICF is a classification of health and health-related domains that helps facilitate a holistic understanding of a particular condition (World Health Organization, 2001). The ICF domains include: body functions and structures; activity; participation; environmental factors; and personal factors. Examining the ICF areas in which sensory subtypes are known to differ allows for a more complete understanding of the needs of each subtype (Figure 1.2). Differences between the subtypes in body functions include the cardiovascular system (i.e., heart rate variability) and auditory perception (i.e., event related potential) (Lane, Eldridge, Hand, Harpster, & Dennis, 2015; Schaaf & Lane, 2015b). Additionally, activities such as communication and daily living skills differ between subtypes (Tanner & Lane, unpublished manuscript). Personal factors refer to influences on functioning that are unique to the individual and are not represented by other ICF domains (World Health Organization, 2001). Personal factors such as maladaptive behaviors, emotional reactivity, nonverbal IQ, attention, and socialization coping skills (i.e., demonstrating responsibility and sensitivity to others) similarly vary from subtype to subtype (Lane et al., 2014a; Tanner & Lane, unpublished manuscript). Given these differences across multiple ICF domains, it is reasonable to hypothesize that the four subtypes may differentially respond to intervention. However, a critical gap in knowledge exists regarding differences between subtypes in environmental factors, which refers to one’s emotional, physical, and attitudinal environment, and participation, which is involvement
in life situations (World Health Organization, 2001). The studies described in Chapters 2 and 3 were designed to address this gap in the existing literature.
Figure 1.2 ICF diagram of work to date on sensory subtypes in ASD

Note. ICF = International Classification of Functioning, Disability, and Health
1.4 Burden Among Caregivers of Children with ASD

Caregivers, including parents, family members, and friends, of individuals with physical, mental, or developmental disabilities often experience negative psychological, behavioral, and physiological effects (Bevans & Sternberg, 2012). These effects are collectively termed caregiver burden and have been widely researched for over four decades (Hoffmann & Mitchell, 1998). Caregiver burden can be conceptualized as an environmental factor, as it is part of the attitudinal and emotional environment of children with ASD. Caregivers are reported to experience both objective burden, which refers to observable, negative events, happenings, and activities, and subjective burden, which refers to feelings, attitudes, and emotions (Montgomery, Gonyea, & Hooyman, 1985).

Subjective burden has been associated with negative health behaviors including: decreased physical activity; poor sleep patterns; difficulties with weight maintenance; smoking; and alcohol consumption (Gallant & Connell, 1998). Moreover, high caregiver burden is associated with poorer quality of life of the caregiver and, as such, may necessitate intervention (Rizk, Pizur-Barnekow, & Darragh, 2014; McCullagh, Brigstocke, Donaldson, & Kalra, 2005).

Caregivers of children with ASD are documented to have higher levels of burden than caregivers of children with attention deficit hyperactive disorder, developmental disabilities, or other healthcare needs (Cadman et al., 2012; Dabrowska & Pisula, 2010; Estes et al., 2009). Parent factors, including coping strategies, difficulty of caregiving tasks, and socioeconomic status, have been linked to the level of caregiver burden (Abbeduto et al., 2004; Lanfranchi & Vianello, 2012; Stuart & McGrew, 2009). Additionally, characteristics of the child, including maladaptive behaviors, physical
health problems, and whether or not the child lives in the home, are associated with the level of burden experienced by caregivers of children with ASD (Kring, Greenberg, & Seltzer, 2008, 2009).

Another child characteristic that is related to level of caregiver burden is sensory features (Kirby, White, & Baranek, 2015; Schaaf, Toth-Cohen, Johnson, Outten, & Benevides, 2011). Specifically, increased levels of hyper- and hypo-reactivity are associated with increased caregiver objective burden, while increased hyper-reactivity alone is associated with increased subjective burden. Sensory features have also been shown to affect burden at the family level, as families of children with ASD report the development of specific strategies that are sensitive to the child’s sensory features for the successful completion of daily activities (Schaaf et al., 2011). To date, literature on the influence of sensory processing on caregiver burden has focused on specific sensory features (hypo- and hyper-reactivity, sensory seeking) and their relationship to caregiver burden, rather than looking at patterns of sensory features common to groups of children with ASD (sensory subtypes). The patterns of sensory features observed among sensory subtypes may include a mixture of hypo-reactivity in one sensory domain and hyper-reactivity in another, for example. Variation in caregiver burden as a function of sensory subtype has yet to be explored.

1.5 Participation of Children with ASD

The value of participation in daily activities in the home, school, workplace, and community is a central tenant of the profession of occupational therapy and is one of the five health-related domains recognized in the ICF (AOTA, 2014; World Health Organization, 2001). The Occupational Therapy Practice Framework and ICF define
participation as active involvement in daily life activities that the individual finds purposeful and meaningful (AOTA, 2014; World Health Organization, 2001).

Participation in valued activities serves as a learning opportunity for children to practice and develop skills and improves quality of life (Dunst, Bruder, Trivette, & Hamby, 2006; Humphry & Wakeford, 2006; The Whoqol Group, 1998).

It is known that children with ASD participate in fewer and less diverse activities than typically developing children and children with other developmental disabilities (LaVesser & Berg, 2011; Potvin, Snider, Prelock, Kehayia, & Wood-Dauphinee, 2012). Within the activities in which children with ASD do participate, they demonstrate significantly less competence than typically developing peers (Reynolds, Bendixen, Lawrence, & Lane, 2011). Child factors, including language ability, autism severity, and cognitive function, have been associated with levels of participation of children with ASD (Little, Ausderau, Sideris, & Baranek, 2015; Orsmond, Shattuck, Cooper, Sterzing, & Anderson, 2013; Rosenberg, Bart, Ratzon, & Jarus, 2012). Further, some studies have documented relationships between sensory features and activity participation in children with ASD (Little et al., 2015). Children with hyper-reactivity have been shown to participate less frequently in activities outside of the home, while those with hypo-reactivity were more likely to participate in activities outside of the home. High levels of sensory interests, repetitions, and seeking behaviors have been associated with increased activity participation within the home, and high levels of enhanced sensory perception are associated with increased frequency of participation in activities across contexts. Additionally, children with increased sensory sensitivity and sensory avoiding, which are behaviors indicative of sensory hyper-reactivity, demonstrated lower participation in
activities, social interaction, and school performance (Reynolds, Bendixen, Lawrence, & Lane, 2011). Taken together, these studies support the notion that sensory features influence activity participation. However, the differences in participation between the four subtypes proposed by Lane and colleagues remain unknown.

1.6 Need for an Intervention Selection Framework

Recent changes in the healthcare field have resulted in increased pressure for clinicians, including occupational therapists, to ensure that intervention choices are based on sound evidence (Bennett & Bennett, 2000). Although occupational therapy is the second most frequently used therapy for children with ASD, a recent survey of occupational therapists revealed that 53% of practitioners lack confidence when working with the ASD population (Ashburner, Rodger, Ziviani, & Jones, 2014). Further, while 93% of respondents identified sensory processing as a key area of knowledge and skill when working with individuals with ASD, 58% found the literature to be inconclusive or contradictory. Respondents also frequently reported clinical reasoning as a priority for future learning, which speaks to the need for clinical decision-making support for occupational therapists working with individuals with ASD.

Findings of the studies described in Chapters 2 and 3 will be combined with existing literature to develop a decision-making framework that links sensory subtypes of children with ASD to known areas of deficit and evidence based interventions. Given the child’s sensory subtype, the decision-making framework will highlight deficits across ICF domains that may be targets for intervention based on research findings. Further, the framework will also suggest evidence-based occupational therapy interventions for each relevant functional deficit. Such a framework has implications for both clinicians, who
may wish to utilize in practice, and researchers, who may wish to test the differential effectiveness of interventions for children in different sensory subtypes. Evidence-based interventions will be drawn from a recent special issue in the American Journal of Occupational Therapy containing four systematic reviews of the effectiveness of occupational therapy interventions for children with ASD (Kuhaneck et al., 2015; Tanner et al., 2015; Watling & Hauer, 2015; Weaver, 2015).

An illustration of how this framework might support clinical reasoning is as follows: A child is determined to best fit in the GSD subtype. The occupational therapist (OT) examines the decision making framework and notices that children in the GSD subtype are known to have more maladaptive behaviors, which are behavioral difficulties that interfere with everyday activities, than children in other subtypes (Lane et al., 2010). The OT then decides to administer an assessment or caregiver interview to determine if their client demonstrates difficulty in this area. Upon confirmation of difficulty in this area, the OT consults the decision-making framework to determine evidence-based interventions that may be helpful for this client. The OT sees that antecedent manipulation and self-management are two behavioral techniques within the scope of occupational therapy that are shown to be effective at reducing maladaptive behaviors (Tanner et al., 2015). The OT then decides to implement these interventions. Guiding clinical reasoning and linking deficits to evidence-based interventions through the use of this novel framework may significantly improve the occupational therapy delivery process for children with ASD.

Similarly, a researcher who wishes to test the effectiveness of a particular intervention may find such a framework useful for developing hypotheses about
differences in outcomes of children in different subtypes. Without the use of such a framework, it is possible that if the researcher did not control for sensory subtype membership, the intervention would incorrectly be deemed ineffective if it benefited children in some subtypes, but not others. Additionally, the use of such a framework may be beneficial in the development and planning of future research studies. For example, it will be important to determine whether or not the use of such a framework for intervention selection ultimately results in improved treatment outcomes for children with ASD.

Currently, the decision-making support for occupational therapists working with children with ASD comes from the Occupational Therapy Practice Guidelines for Individuals with Autism Spectrum Disorder (Tomchek & Koenig, 2016). This practice guideline “provides an overview of the evidence for occupational therapy to assist people with autism in achieving health, well-being, and participation.” The decision-making framework proposed in Chapter 5 differs from this document in that it walks clinicians and researchers through a process starting with determining the child’s sensory subtype, to known areas of difficulty for that subtype, then to evidence-based interventions that may be effective for that child. The current practice guidelines provide a summary of evidence-based interventions for individuals with ASD as a whole, but are not customized to subgroups within ASD. As such, they may be used as a complement to the sensory subtype-based decision making framework. The framework proposed in Chapter 5 offers a unique solution to the phenotypic heterogeneity in ASD that complicates intervention selection by customizing clinical reasoning for each homogenous sensory subtype. By narrowing the scope of intervention for each subtype and linking functional
deficits with evidence-based practices, this framework has the potential to significantly improve the quality of care for children with ASD.

### 1.6.1 Clinical and research utility

An important factor in the development of decision-making supports is efficiency and utility. Indeed, research demonstrates that aspects of clinical utility including ease of use and administration time are of concern to clinicians who may wish to utilize such a decision-making framework in their daily practice (Smart, 2006). Additionally, there is an existing body of literature that suggests that reduced research survey length is associated with improved response rate, higher quality of responses, and more complete surveys (Deutskens, Ruyter, Wetzel, & Oosterveld, 2004; Fan & Yan, 2010; Haunberger, 2011). Currently, the algorithm that is used to determine a child’s sensory subtype membership is based on SSP domain-level z-scores, requiring caregivers to answer all 38 items. However, short forms consisting of reduced item subsets have become increasingly popular with the funding of the Patient Reported Outcomes Measures Information System (PROMIS) by the National Institutes of Health (NIH). Preliminary work in our lab indicates that not all items from the SSP are salient to subtype determination (Hand, Lane, Marco, & Boeck, 2016). As such, a reduction in the number of items may be possible. Reducing the number of items for subtype determination will increase the efficiency of measurement and reduce respondent burden, which may be beneficial during evaluations and research studies where time is limited.

### 1.8 Objectives

There is significant relevance of sensory subtyping to diagnosis, intervention selection, and intervention efficacy for children with ASD. Indeed, differences between...
sensory subtypes have been documented in many areas of function, suggesting that intervention efficacy may vary by subtype (Lane et al., 2010, 2014a; Tanner & Lane, unpublished manuscript). There is a paucity of literature, however, that examines differences between sensory subtypes in caregiver burden and child participation. This gap in knowledge provides an opportunity to investigate this novel area of functioning among the subtypes. Successful findings could improve treatment outcomes for children with ASD by providing a clinical decision making framework that links functional performance limitations of each sensory subtype, across ICF health-related domains, to evidence-based interventions. Further, preliminary work in our lab indicates that a reduction in the number of items needed to determine sensory subtype may be possible, which will increase the efficiency of measurement and utility in clinical and research settings, while reducing respondent burden (Hand et al., 2016).

The long-term goal of this project is to improve the clinical and research utility of sensory subtyping by determining differences in caregiver burden and participation between the subtypes and by refining the classification mechanism. The following objectives are designed to achieve this goal:

1. Examine caregiver burden as a function of sensory subtype
2. Examine child participation in home, school, and community activities as a function of sensory subtype
3. Determine if a reduced item subset could be used to reliably identify sensory subtypes
1.9 Research Aims

**Aim 1: Determine differences in caregiver burden between sensory subtypes of children with ASD.**

**Hypothesis 1:** Caregivers of children in subtypes with more severe sensory features will report greater caregiver burden than those with less severe sensory features. In other words, caregivers of children in the GSD subtype will report the highest levels of burden, caregivers of children in the TSS and PI subtypes will report intermediate levels of burden, and caregivers of children in the SA subtype will report mild levels of burden.

**Aim 2: Determine differences in participation between sensory subtypes of children with ASD.**

**Hypothesis 2:** Children in subtypes with mild sensory features will have greater participation in home, school, and community activities than those with more severe sensory features. Specifically, children in the SA subtype will display the highest levels of participation, while children in the TSS and PI subtypes will demonstrate intermediate participation, and children in the GSD subtype will be associated with the most limited participation.

**Aim 3: Establish a subset of SSP items that identify sensory subtypes of children with ASD.**

**Hypothesis 3:** A subset of 4-10 SSP items can identify a child’s sensory subtype with >90% agreement with the current gold standard.
1.10 Summary

Completion of this series of studies will result in areas of caregiver burden and child participation that differ between sensory subtypes of children with ASD. The results will improve the clinical utility of sensory subtyping by linking subtypes to areas of caregiver burden and child participation to target for intervention. Study findings, in conjunction with previously documented differences between the subtypes, can be used to create a clinical decision making framework to focus the scope of intervention for each subtype, which may subsequently improve treatment outcomes for children with ASD. Additionally, by reducing the number of questions that caregivers must answer to determine a child’s sensory subtype will improve the clinical utility of sensory subtyping and reduce respondent burden.
Chapter 2: Caregiver Burden as a Function of Sensory Subtypes of Children with Autism

2.1 Abstract

Background: Level of burden experienced by caregivers of children with autism spectrum disorder (ASD) has been shown to be a function of many parent and child characteristics, including the child’s difficulty with reacting to everyday environmental sensory stimuli. Recently, researchers have proposed a classification schema for children with ASD based on shared patterns of sensory difficulties. This schema groups children with ASD into one of four sensory subtypes: Sensory Adaptive; Taste/Smell Sensitive; Postural Inattentive; and Generalized Sensory Difference. However, variation in the level of burden experienced by caregivers as a function of the child’s sensory subtype has yet to be explored. Purpose: The purpose of the present study is to examine the relationship between child sensory subtype and the level of burden experienced by the caregiver. Two aspects of caregiver burden were examined in this study: 1) health-related quality of life (HRQOL) and 2) caregiver strain. Method: A national online survey of caregivers of children with ASD was conducted (n = 367). Canonical correlation analyses were used to examine the relationship between child sensory subtype and indicators of caregiver burden. Results: The analyses revealed that there was not a significant association between the child’s sensory subtype and caregiver reported HRQOL. Caregiver strain was significantly associated with the child’s sensory subtype membership such that, when
compared to caregivers of children in the Generalized Sensory Difference subtype, caregivers of children in the Sensory Adaptive subtype reported the lowest levels of caregiver strain, followed by caregivers of children in the Taste/Smell Sensitive subtype. Caregivers of children in the Postural Inattentive and Generalized Sensory Difference subtypes did not significantly differ in levels of caregiver strain. Older age of the caregiver and child, as well as higher household income, were associated with lower caregiver strain. Conclusions: This study demonstrates that strain of caregivers of children with ASD is associated with the child’s sensory subtype. Possible mechanisms that may explain the relationship between caregiver strain and child sensory subtype membership are discussed.
2.2 Introduction

It is widely documented that caregivers of children with autism spectrum disorder (ASD) have higher levels of burden than caregivers of children with attention deficit hyperactive disorder, developmental disabilities, or other healthcare needs (Cadman et al., 2012; Dabrowska & Pisula, 2010; Estes et al., 2009). Two common ways of quantifying caregiver burden are to examine the level of health related quality of life (HRQOL) and strain of the caregiver (Khanna et al., 2011). HRQOL is a measure of “perceived mental and physical health over time,” and captures ways in which mental and physical health influence quality of life, while caregiver strain reflects “the demands, responsibilities, difficulties, and negative psychic consequences of caring for relatives with special needs” (Centers for Disease Control (CDC), 2016; Brannan, Heflinger, & Bickman, 1997). Poor HRQOL has been associated with chronic diseases and chronic disease risk factors, including high body mass index, physical inactivity, and smoking (CDC, 2016). Similarly, high caregiver strain has been linked with negative health behaviors including: decreased physical activity; poor sleep patterns; difficulties with weight maintenance; smoking, and alcohol consumption (Gallant & Connell, 1998).

Given the long-term implications of poor HRQOL and caregiver strain, it is critical to understand the factors associated with burden in caregivers of children with ASD.

Various parent and child characteristics are associated with the level of HRQOL and strain experienced by caregivers of children with ASD. Parent factors, such as higher socioeconomic status, more perceived social support, and adaptive coping strategies, are associated with better HRQOL in caregivers of children with ASD (Lee et al., 2009; Khanna et al., 2011). Lower levels of social support, negative cognitive appraisal of
situations, and passive avoidant coping strategies (i.e., denial, avoidance) have been associated with increased caregiver strain (Stuart & McGrew, 2009). Characteristics of the child, such as maladaptive behaviors, physical health problems, and whether or not the child lives in the home, are related to caregiver HRQOL, while severity of ASD symptoms, level of behavioral difficulties, and functional impairment have been linked with caregiver strain (Kring et al., 2008, 2009; Khanna et al., 2012; Stuart & McGrew, 2009).

Sensory features, which refer to behaviors associated with difficulties processing and integrating sensory information, are another child factor that is associated with caregiver HRQOL and strain (Kirby, White, & Baranek, 2015; Schaaf & Lane, 2015; Schaaf, Toth-Cohen, Johnson, Outten, & Benevides, 2011). Individuals with ASD and sensory features may: a) demonstrate hyper- or hypo-reactivity to sensory stimuli; b) have unusual sensory interests; c) inaccurately perceive sensory stimuli; and/or d) have difficulties managing multiple concurrent sensory stimuli (Schaaf & Lane, 2015a). Recent work indicates that increased levels of hyper- and hypo-responsiveness are associated with increased caregiver objective burden, and hyper-responsiveness is also significantly associated with increased caregiver subjective burden (Kirby et al., 2015).

Recently, researchers have found that children with ASD can be classified into sensory subtypes, which are groups of children who demonstrate shared patterns of sensory features (Ausderau et al., 2014; Lane et al., 2010, 2011, 2014a). There are multiple potential advantages to the use of sensory subtypes including the ability to narrow the scope of intervention by selecting those interventions that are likely to be the most effective for children in each sensory subtype. The earliest, and most extensively
researched sensory subtypes, are those proposed by Lane and colleagues (Lane et al., 2010, 2011, 2014a). Lane et al. (2014a) propose a classification schema of sensory subtypes of children with ASD based on parent responses to the Short Sensory Profile (SSP; McIntosh, Miller, Shyu, & Dunn, 1999). Using this schema, four sensory subtypes in ASD have been identified: 1) Sensory Adaptive; 2) Taste/Smell Sensitive; 3) Postural Inattentive; and 4) Generalized Sensory Difference (Lane et al., 2014a). The four subtypes differ by severity of difficulties displayed in the SSP domains of under-responsive/seeks sensation and auditory filtering, with Sensory Adaptive (SA) representing the least severe subtype and Generalized Sensory Difference (GSD), the most. While the SA subtype is characterized by mild disturbance in the SSP domains of auditory filtering and under-responsive/seeks sensation, it is unlikely that functional performance is strongly affected by these sensory features. The two intermediate severity subtypes, Taste/Smell Sensitive (TSS) and Postural Inattentive (PI), are characterized by extreme taste/smell sensitivity and difficulties with maintaining postural integrity, respectively. The GSD subtype demonstrates clinically meaningful difficulties in all seven SSP sensory domains.

Distinct non-sensory behavior profiles have also been associated with each sensory subtype. For example, children in the TSS subtype display increased picky eating behaviors and more severe communication impairments, while children in the GSD subtype demonstrate more maladaptive behaviors (Lane et al., 2010, 2011). Additionally, differences in daily living skills, emotional regulation, and attention between the subtypes have been documented (Tanner & Lane, unpublished manuscript). Although the sensory subtypes differ from one another in clinical presentation, they are not well explained by
systematic variation in age, non-verbal IQ, or severity of autistic symptoms including social interaction, communication, restricted and repetitive behaviors (Lane et al., 2014a).

As the sensory subtypes are documented to differ in various personal characteristics, behaviors, and activities, it is reasonable to hypothesize that caregivers of children in different subtypes experience different levels of caregiver burden. Additional information about the level of caregiver burden as a function of subtype may be useful in developing much-needed clinical reasoning supports for clinicians working with children with ASD by identifying potential aspects of burden in need of intervention for each sensory subtype (Ashburner et al., 2014). However, to date, the level of caregiver burden experienced as a function of sensory subtype has not been explored. As a next step toward the widespread clinical use of sensory subtyping to guide intervention selection for children with ASD and their caregivers, the objective of the present study is to examine level of caregiver burden as a function of sensory subtype. Specifically, we will:

1. Determine differences between sensory subtypes of children with ASD on caregiver HRQOL; and
2. Determine differences between sensory subtypes of children with ASD on caregiver strain.

Our hypothesis for objective 1 is that caregiver HRQOL will be inversely related to level of severity of sensory features. Specifically, caregivers of children in the least severe subtype (SA) will report the highest HRQOL, while caregivers of children in the most severe subtype (GSD) will report the lowest HRQOL. Caregivers of children in the intermediate severity subtypes (TSS and PI) are hypothesized to report levels of HRQOL somewhere between that of caregivers of children in the mild and severe subtypes. Our
hypothesis for objective 2 is that caregiver strain is directly proportional to level of severity of sensory features, in that caregivers of children in the GSD subtype will report the highest levels of strain, followed by caregivers of children in the TSS and PI subtypes. Caregivers of children in the SA subtype are hypothesized to report the lowest levels of strain.

2.3 Materials and Methods

2.3.1 Study Design

The present study used a non-experimental, cross-sectional design. The primary variables of interest were: 1) caregiver burden, which includes caregiver HRQOL, as measured by the Short Form 12 Health Survey version 2 (Ware, Kosinski, & Keller, 1996), and caregiver strain, as measured by the Caregiver Strain Questionnaire (Brannan et al., 1997), and 2) sensory subtype of the child for whom the participant provides care, as measured by the Short Sensory Profile. Age of the caregiver, age of the child, household income, and number of children in the household were included as covariates due to the influence that these variables have on caregiver burden in other populations (Goldstein et al., 2004; Hastings, 2002; Liu, Lambert, & Lambert, 2007; Nabors, Seacat, & Rosenthal, 2002).

2.3.2 Participants and Procedures

Participants of the present study were primary caregivers of children aged 5-13 years with ASD (n = 367). For the purpose of this study, a primary caregiver is the person who takes primary responsibility for the child with ASD. To eliminate potential confounding effects on level of caregiver burden, caregivers were excluded from the present study if their child had a comorbid: 1) neurological diagnosis (i.e., cerebral palsy,
stroke, spinal cord injury); 2) genetic diagnosis (i.e., Rett, Down, or Fragile X syndromes); and/or 3) a significant physical disability (i.e., blindness or deafness).

Participants were recruited through multiple avenues including the Interactive Autism Network (IAN), ResearchMatch, and pediatric clinics in the greater Columbus, Ohio area. The IAN is an online research registry for caregivers of children with ASD, which recently authenticated the parent-report ASD diagnoses of children in their database via medical record review (Daniels et al., 2011), and ResearchMatch is a national online registry for individuals interested in being matched with ongoing studies for which they qualify. A total of 8,215 and 255 registrants met our inclusion criteria from the IAN and ResearchMatch, respectively. Participants meeting study criteria from the IAN and ResearchMatch received a brief study description with a link to the survey site via email. Three total emails were sent to each potential participant to maximize survey response rate. Additionally, fliers for the present study were posted in local pediatric occupational, physical, and speech therapy clinic waiting rooms and reception areas. Participants received a free Redbox DVD code via email for survey completion. Figure 2.1 provides a schematic representation of the survey response rate and retention throughout completion of the online survey. This study was reviewed and approved by the Institutional Review Board of The Ohio State University.
Figure 2.1 Caregiver burden survey response rate

Note. SF-12v2 = Short Form 12 Health Survey, version 2; SSP = Short Sensory Profile; CGSQ = Caregiver Strain Questionnaire
2.3.3 Measures

*Caregiver Health-Related Quality of Life.*

The Short Form 12 Health Survey version 2 (SF-12v2) was used as a measure of caregiver HRQOL (Ware, Kosinski, & Keller, 1996). The SF-12v2 is a widely utilized questionnaire in survey research, including studies seeking to measure the HRQOL of caregivers of children with ASD (Khanna et al., 2011). This questionnaire is a 12-item, norm-referenced, self-report measure that assesses eight health domains that combine to yield aggregate summary measures for Physical Component Summary (PCS) and Mental Component Summary (MCS), where higher scores indicate better health. The PCS and MCS have been shown to have excellent internal consistency in the general population with reliability coefficients (Cronbach’s $\alpha$) of 0.92 and 0.88, respectively (Maruish, 2012).

*Caregiver Strain.*

Level of caregiver strain was assessed with the Caregiver Strain Questionnaire (CGSQ, Brannan et al., 1997). The CGSQ is a 21-item questionnaire that measures three types of strain: 1) objective strain, which is negative happenings or events as a result of the child’s behavioral difficulties; 2) subjective internalized strain, which refers to negative feelings experienced by the caregiver; and 3) subjective externalized strain, which is negative feelings that the caregiver has towards the individual for whom they provide care (Kirby et al., 2015). The CGSQ has been shown to have excellent internal consistency reliability with Cronbach’s $\alpha = 0.93$ (Brannan et al., 1997). While the CGSQ was originally developed for caregivers of children with emotional and behavioral difficulties, it has been validated for use in the population of caregivers of children with
ASD (Brannan et al., 1997; Khanna et al., 2012; Stuart & McGrew, 2009). Within the population of caregivers of children with ASD, the CGSQ demonstrates excellent internal consistency reliability (Cronbach’s $\alpha = 0.94$), and has been shown to have convergent validity with conceptually similar measures (Khanna et al., 2011; Khanna et al., 2012). Further, the three-construct (objective, subjective internalized, and subjective externalized strain) factor structure was validated in a sample of caregivers of children with ASD (Khanna et al., 2012).

*Child Sensory Subtype.*

The Short Sensory Profile was administered for the purpose of determining child sensory subtype (McIntosh et al., 1999). The SSP is a parent questionnaire consisting of 38 items that are designed to measure behaviors associated with abnormal sensory processing. The SSP evaluates sensory processing in seven sensory domains: 1) tactile; 2) taste/smell; 3) movement; 4) visual/auditory sensitivity; 5) under-responsive/seeks sensation; 6) auditory filtering; and 7) low energy/weak. Scores for the seven sensory domains are compared to normative data from 1,200 typically developing children. Higher scores indicate a more typical performance, while lower scores indicate a probable or definite difference in sensory processing. Internal consistency of overall and subdomain sections of the SSP is moderate to excellent ($r = 0.70 - 0.90$) and discriminative validity is acceptable (> 95% in differentiating children with and without sensory impairments, McIntosh et al., 1999). While the SSP was originally designed and validated for measuring sensory processing in children aged 3-10 years, it has been widely used in populations of children over 10 years of age (Mangeot et al., 2001; Schoen, Miller, & Green, 2008; Tavassoli et al., 2016; Uljarević, Lane, Kelly, & Leekam, 2016).
Moreover, the SSP demonstrates convergent validity with conceptually similar measures in individuals with ASD up to 14 years of age (Tavassoli et al., 2016), and in individuals with sensory processing difficulties, but not ASD, up to 16 years of age (Schoen et al., 2008). As such, a broader age range of participants is reflected in the present sample.

2.3.4 Data analyses

Participants who had missing data rates greater than 15% for the SF-12v2 (n = 2) or the SSP (n=3) were excluded from analyses. No participants had missing data rates greater than 15% for the CGSQ. Additionally, participants who did not report child age (n = 3) were excluded, as their eligibility for the study could not be confirmed. Lastly, participants who did not complete 50% or more items from any single SSP domain (n = 3) were excluded, due to the impact this may have on subtype determination.

Missing data rates for the remaining participants (n = 367) were as follows: 0.49% for the SF-12v2, 0.42% for the CGSQ, and 0.24% for the SSP.

Iterative model-based imputation using robust stepwise regression was used to impute any missing values for the remaining 367 participants with package “VIM” in the statistical software R (Templ, Alfons, Kowarik, Prantner, & Templ, 2015). With this imputation approach, in each step of the iteration, one variable is used as a dependent variable and the remaining variables serve as the independent variables in a regression model. Demographic information was summarized descriptively and sensory subtypes for each child were determined from caregiver responses to the SSP using the algorithm proposed by Lane and colleagues.
As an extra validation procedure, because the impact of imputing missing SSP values on sensory subtype determination has not yet been tested, a content area expert on sensory subtypes examined SSP domain z-scores for participants with missing values (n = 25) prior to imputation. Based on visual inspection of z-scores, the expert identified the subtype in which they hypothesized each participant belonged. The agreement rate between the classifications based on the expert’s visual inspection and the subtype classification based on the imputed values was 68%. While this is a relatively conservative level of agreement, we chose to include these participants in subsequent analyses, as the missing data rate overall for the SSP was small (0.24%).

PCS and MCS scores were calculated from SF-12v2 data using the SF-12v2 Scoring Software 5.0. Participant factor scores for objective, subjective internalized, and subjective externalized strain were extracted from the CGSQ using the confirmatory factor analysis model published by Khanna et al. (2012) in the statistical software R using package “sem” (Fox et al., 2016). Lastly, canonical correlation analysis (CCA), a multivariate generalized linear modeling technique, was conducted to explore the relationship between caregiver burden and person characteristics, including the child’s sensory subtype.

Canonical correlation analysis can be conceptualized as “a logical extension of multiple regression” (Hair, 2006). While multiple regression allows for examination of a single dependent variable and several independent variables, CCA allows for the concurrent examination of several dependent variables and several independent variables. CCA generates a linear combination of each set of variables (independent and dependent) that maximizes the correlation while the variance for each variable that is attributable to
the other variables in the set is controlled (Hair, 2006; Dardas & Ahmad, 2014). These linear combinations, called canonical functions, can be interpreted like multiple regression equations (Sherry & Henson, 2005). The number of canonical functions generated for a given model is equal to the number of variables in the smaller of the two variable sets. Each canonical function yields a canonical correlation, which is a measure of the relationship between the independent and dependent variables and can be conceptualized as a Pearson correlation (Sherry & Henson, 2005).

CCA provides multiple advantages over univariate statistical analyses. First, CCA limits the probability of Type I error due to the multivariate nature of the analysis (Fish, 1988). Additionally, as caregiver HRQOL and strain are complex constructs that are influenced by multiple variables and the interaction of those variables, this technique allows for the detection of important multivariate relationships that may not be captured when using univariate methods.

In the present study, two CCA’s were performed. The first analysis examined the relationship between a set of personal characteristics (child sensory subtype, child age, caregiver age, household income, number of children in the household) and a set of caregiver HRQOL variables (PCS, MCS). The second analysis examined the relationship between the same personal characteristics and a set of caregiver strain variables (objective, subjective internalized, subjective externalized). A dummy-coding scheme was used for the categorical variable of sensory subtype, with GSD as the reference group; dummy coding uses ones and zeros to convey information about group membership and creates \( k-1 \) new variables, where \( k \) = the number of groups, to represent a categorical variable. Table 2.1 provides a visual representation of the dummy-coding
scheme used in this study. Figures 2.2a and 2.2b provide graphic illustrations of the CCAIs conducted in the present study. Both CCA analyses were conducted using the statistical software program R with ad-on package “yacca” (R Development Core Team, 2011; Butts & Butts, 2009).
Table 2.1 Visual representation of dummy coding for sensory subtype membership

<table>
<thead>
<tr>
<th>Subtype</th>
<th>V1</th>
<th>V2</th>
<th>V3</th>
</tr>
</thead>
<tbody>
<tr>
<td>SA</td>
<td>1</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>TSS</td>
<td>0</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>PI</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>GSD</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
</tbody>
</table>

Note. SA = Sensory Adaptive; TSS = Taste/Smell Sensitive; PI = Postural Inattentive; GSD = Generalized Sensory Difference; V1, V2, and V3 represent the dummy variables.
Canonical correlations were interpreted using a multi-step procedure (Sherry & Henson, 2005). First, the overall significance of the full model was evaluated via significance test of Wilk’s lambda ($\lambda$). Second, the relative importance of individual functions was evaluated by examining Wilk’s $\lambda$ and squared canonical correlations ($R^2_c$), which represent the variance-accounted-for effect size and are analogous to $R^2$ effects in multiple regression. Third, for significant functions, the importance of individual variables were interpreted using the following values: 1) standardized canonical coefficients, which are the coefficients used in the linear equations to combine the observed independent and dependent variables into respective synthetic variables; 2) structural correlations, which are bivariate correlations between the observed and synthetic variables; and 3) squared structural correlations, which indicate the amount of variance an observed variable linearly shares with its respective synthetic variable (Sherry & Henson, 2005). A cutoff value of 0.30 was used to determine significance of structural correlations (Dardas & Ahmad, 2014; Matson, Neal, Fodstad, & Hess, 2010).
Figure 2.2 Schematic representations of canonical correlation analyses for caregiver burden and personal characteristics

2.2a Analysis of caregiver HRQOL and personal characteristics

*Child sensory subtype variable; SA = Sensory Adaptive; TSS = Taste/Smell Sensitive; PI = Postural Inattentive; GSD = Generalized Sensory Difference (reference group); PCS = Physical composite summary; MCS = Mental composite summary

Continued on next page
2.2b Analysis of caregiver strain and personal characteristics

*Child sensory subtype variable; SA = Sensory Adaptive; TSS = Taste/Smell Sensitive; PI = Postural Inattentive; GSD = Generalized Sensory Difference (reference group)
Power analysis.

An a-priori power analysis was conducted using G*Power 3.1 for a Pearson’s $r$ with a small (Cohen’s $\eta^2 = 0.30$) effect size at 80% power (Faul, Erdfelder, Buchner, & Lang, 2009; Dardas & Ahmad, 2014). The power analysis yielded a necessary sample size of 282.

2.4 Results

A total of 367 usable survey responses were received. Table 2.2 provides descriptive information about the participants. On average, respondents were 42.46 years of age (SD = 6.87 years), and 83.38% were female. Over 82% of the respondents were married. Nearly 69% of participants were employed full or part time and 73% had an associate’s degree or higher. The majority of children with ASD for whom the participants of the present study provide care were male (93.73%), with an average age of 10.09 years (SD = 2.51 years). Descriptive statistics for participant scores on the SF-12v2 and CGSQ for all participants and by subtype are detailed in Table 2.3. Bivariate correlations for person characteristic variables and measures of caregiver burden are presented in Table 2.4.
Table 2.2 Caregiver characteristics

<table>
<thead>
<tr>
<th></th>
<th>All</th>
<th>SA</th>
<th>TSS</th>
<th>PI</th>
<th>GSD</th>
</tr>
</thead>
<tbody>
<tr>
<td>n</td>
<td>367</td>
<td>20</td>
<td>137</td>
<td>77</td>
<td>133</td>
</tr>
<tr>
<td>Mean age (SD)</td>
<td>42.46 (6.87)</td>
<td>43.33 (3.91)</td>
<td>42.95 (7.12)</td>
<td>41.82 (6.42)</td>
<td>42.14 (7.22)</td>
</tr>
<tr>
<td>Age range</td>
<td>25.82-66.80</td>
<td>36.20-50.02</td>
<td>28.74-66.80</td>
<td>25.82-61.28</td>
<td>29.08-61.18</td>
</tr>
<tr>
<td>% Female</td>
<td>83.38</td>
<td>75.00</td>
<td>83.94</td>
<td>81.82</td>
<td>84.97</td>
</tr>
<tr>
<td>Household income*</td>
<td>100k+</td>
<td>100k+</td>
<td>100k+</td>
<td>100k+</td>
<td>25-50k</td>
</tr>
<tr>
<td>Median number of children (range)</td>
<td>2 (1-9)</td>
<td>2 (1-9)</td>
<td>2 (1-5)</td>
<td>2 (1-6)</td>
<td>2 (1-5)</td>
</tr>
<tr>
<td>Highest level of education*</td>
<td>Some graduate school</td>
<td>Associates degree</td>
<td>Some graduate school</td>
<td>Some graduate school</td>
<td>Some college/Associates degree</td>
</tr>
<tr>
<td>Employment status*</td>
<td>Full time</td>
<td>Part time</td>
<td>Part time</td>
<td>Full time</td>
<td>Full time</td>
</tr>
<tr>
<td>Marital status*</td>
<td>Married</td>
<td>Married</td>
<td>Married</td>
<td>Married</td>
<td>Married</td>
</tr>
<tr>
<td>Mean child’s age (SD)</td>
<td>10.09 (2.51)</td>
<td>10.19 (2.57)</td>
<td>9.95 (2.67)</td>
<td>10.29 (2.50)</td>
<td>10.11 (2.35)</td>
</tr>
</tbody>
</table>

Note. *Mode. All ages are listed in years. SD = Standard deviation; SA = Sensory Adaptive; TSS = Taste/Smell Sensitive; PI = Postural Inattentive; GSD = Generalized Sensory Difference
Table 2.3 Descriptions of SF-12v2 and CGSQ scores

<table>
<thead>
<tr>
<th></th>
<th>All</th>
<th>SA</th>
<th>TSS</th>
<th>PI</th>
<th>GSD</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>SF-12v2 (Scoring Range 0 – 100)</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>PCS Mean</td>
<td>54.15</td>
<td>55.81</td>
<td>54.93</td>
<td>53.24</td>
<td>53.62</td>
</tr>
<tr>
<td>PCS Median</td>
<td>56.46</td>
<td>56.72</td>
<td>58.10</td>
<td>55.10</td>
<td>56.20</td>
</tr>
<tr>
<td>PCS Range</td>
<td>21.61-69.27</td>
<td>38.44-67.73</td>
<td>30.28-69.27</td>
<td>25.10-66.29</td>
<td>21.61-66.58</td>
</tr>
<tr>
<td>PCS IQR</td>
<td>49.38-59.90</td>
<td>53.64-59.95</td>
<td>51.40-59.94</td>
<td>48.81-58.48</td>
<td>48.69-59.86</td>
</tr>
<tr>
<td>MCS Mean</td>
<td>41.07</td>
<td>42.81</td>
<td>41.60</td>
<td>41.77</td>
<td>39.87</td>
</tr>
<tr>
<td>MCS Median</td>
<td>40.34</td>
<td>41.37</td>
<td>41.60</td>
<td>39.69</td>
<td>39.95</td>
</tr>
<tr>
<td>MCS Range</td>
<td>13.76-62.73</td>
<td>27.10-62.38</td>
<td>13.76-62.73</td>
<td>20.05-61.46</td>
<td>15.41-60.54</td>
</tr>
<tr>
<td>MCS IQR</td>
<td>33.46-62.73</td>
<td>35.72-62.73</td>
<td>34.82-62.73</td>
<td>32.89-51.18</td>
<td>32.13-48.83</td>
</tr>
<tr>
<td><strong>CGSQ (Scoring Range -3.0 – +3.0)</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Objective Mean</td>
<td>0.00</td>
<td>-0.90</td>
<td>-0.23</td>
<td>-0.02</td>
<td>0.40</td>
</tr>
<tr>
<td>Objective Median</td>
<td>0.01</td>
<td>-1.05</td>
<td>-0.26</td>
<td>-0.13</td>
<td>0.48</td>
</tr>
<tr>
<td>Objective Range</td>
<td>-1.84-1.98</td>
<td>-1.84-0.87</td>
<td>-1.70-1.98</td>
<td>-1.69-1.90</td>
<td>-1.59-1.97</td>
</tr>
<tr>
<td>Objective IQR</td>
<td>-0.75-0.75</td>
<td>-1.39-(-0.60)</td>
<td>-0.87-0.36</td>
<td>-0.78-0.64</td>
<td>-0.25-1.13</td>
</tr>
<tr>
<td>Subjective int. Mean</td>
<td>0.00</td>
<td>-0.69</td>
<td>-0.18</td>
<td>0.02</td>
<td>0.28</td>
</tr>
<tr>
<td>Subjective int. Median</td>
<td>0.03</td>
<td>-0.72</td>
<td>-0.19</td>
<td>-0.06</td>
<td>0.46</td>
</tr>
<tr>
<td>Subjective int. Range</td>
<td>-1.80-1.56</td>
<td>-1.80-0.90</td>
<td>-1.65-1.48</td>
<td>-1.51-1.53</td>
<td>-1.71-1.56</td>
</tr>
<tr>
<td>Subjective int. IQR</td>
<td>-0.60-0.60</td>
<td>-1.20-(-0.35)</td>
<td>-0.72-0.41</td>
<td>-0.54-0.58</td>
<td>-0.12-0.82</td>
</tr>
<tr>
<td>Subjective ext. Mean</td>
<td>0.00</td>
<td>-0.34</td>
<td>-0.13</td>
<td>0.04</td>
<td>0.15</td>
</tr>
<tr>
<td>Subjective ext. Median</td>
<td>-0.12</td>
<td>-0.43</td>
<td>-0.22</td>
<td>-0.16</td>
<td>0.06</td>
</tr>
<tr>
<td>Subjective ext. Range</td>
<td>-1.01-1.84</td>
<td>-1.01-1.84</td>
<td>-0.98-2.13</td>
<td>-0.85-1.67</td>
<td>-0.95-2.09</td>
</tr>
<tr>
<td>Subjective ext. IQR</td>
<td>-0.52-0.39</td>
<td>-0.74-(-0.15)</td>
<td>-0.57-0.21</td>
<td>-0.46-0.41</td>
<td>-0.37-0.49</td>
</tr>
</tbody>
</table>

Note. SA = Sensory Adaptive; TSS = Taste/Smell Sensitive; PI = Postural Inattentive; GSD = Generalized Sensory Difference; IQR = Interquartile range, presented as upper – lower; PCS = Physical Composite Summary; MCS = Mental Composite Summary; Subjective int. = Subjective internalized strain; Subjective ext. = Subjective externalized strain
Table 2.4 Bivariate correlations between person characteristics and caregiver burden variables

<table>
<thead>
<tr>
<th></th>
<th>HRQOL</th>
<th>Strain</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>PCS</td>
<td>MCS</td>
</tr>
<tr>
<td>SA (vs. GSD)</td>
<td>0.05</td>
<td>0.04</td>
</tr>
<tr>
<td>TSS (vs. GSD)</td>
<td>0.07</td>
<td>0.04</td>
</tr>
<tr>
<td>PI (vs. GSD)</td>
<td>-0.06</td>
<td>0.03</td>
</tr>
<tr>
<td>Age</td>
<td>-0.02</td>
<td>0.03</td>
</tr>
<tr>
<td>Child age</td>
<td>-0.02</td>
<td>0.08</td>
</tr>
<tr>
<td>Household income</td>
<td>0.06</td>
<td>0.06</td>
</tr>
<tr>
<td>Number of children</td>
<td>0.03</td>
<td>0.04</td>
</tr>
</tbody>
</table>

*Note.* *p < 0.05; †p ≤ 0.01; PCS = Physical Composite Summary; MCS = Mental Composite Summary; SA = Sensory Adaptive; TSS = Taste/Smell Sensitive; PI = Postural Inattentive; GSD = Generalized Sensory Difference
All CCA assumptions were examined and confirmed, indicating that the analysis meets criteria for significance testing (Tabachnick & Fidell, 2001). The assumption of linearity was met for all variables within and across variable sets, based on visual inspection of scatterplots. All variable pairs within and across variable sets were evaluated via Levene’s test and were found to meet the assumption of homoscedasticity. The assumption of normality was examined descriptively and all variables, with the exception of number of children, were reasonably normally distributed upon visual inspection. Number of children in the household was found to be positively skewed; however, as the impact of non-normality on multivariate analyses effectively diminishes with sample sizes of at least 200, the present analysis is suitable for interpretation (Hair, 2006, p. 86; Tabachnick & Fidell, 2001, p. 74-75). Although it is not a requirement for CCA, the absence of multicollinearity (i.e., correlations ≥ |0.90|) was evaluated (Nimon, Henson, & Gates, 2010). Examining the bivariate correlations within and between the person and participation variable sets revealed an absence of multicollinearity in all variables with the exception of objective and subjective internalized strain ($r = 0.94$). All within-set correlations were $r \leq 0.54$ and remaining between-set correlations were $r \leq 0.25$.

### 2.4.1 Caregiver HRQOL as a function of sensory subtype

Collectively, the full model was not statistically significant (Wilk’s $\lambda = 0.96$, $\chi^2(14) = 15.76$, $p > 0.05$). As a result, the two canonical functions generated are not suitable for interpretation (Sherry & Henson, 2005). Thus, the present analysis did not detect a relationship between PCS and MCS scores from the SF-12v2 and the person characteristics included in this model.
2.4.2 Caregiver strain as a function of sensory subtype

Collectively, the full model was statistically significant (Wilk’s $\lambda = 0.76, \chi^2(21) = 96.38, p < 0.001$). Evaluation of the $R^2_c$ revealed that the full model explained 24.2% of the variance shared between the two variable sets ($R^2_c = 0.242$). Function 1 accounted for over 17% of the variance explained ($R^2_c = 0.177$). While Function 2, when tested in isolation, was statistically significant (Wilk’s $\lambda = 0.93, \chi^2(12) = 26.23, p < 0.01$), it accounted for less than 5% of the variance explained ($R^2_c = 0.049$), and thus was not practically significant. As a result, Function 2 was not considered in subsequent interpretation. Function 3 did not explain a significant amount ($R^2_c = 0.022$) of shared variance between the two variable sets (Wilk’s $\lambda = 0.98, \chi^2(5) = 8.09, p > 0.05$).

Results of the first canonical function (Table 2.5) indicate that all caregiver strain variables had significant structural correlations, however the effect size was highest for objective ($r_s^2 = 0.98$) and subjective externalized strain ($r_s^2 = 0.77$). The high structural correlation ($r_s = -0.58$) but low standardized coefficient (coef = 0.08) and comparatively low effect size ($r_s^2 = 0.34$) of subjective internalized strain is explained by the previously noted multicollinearity between this variable and objective strain. The significant variables from the person characteristics set were SA, TSS, caregiver age, child age, and household income. Examination of the squared structural correlations reveals that SA and TSS had the largest effect sizes of the person characteristic variables ($r_s^2 = 0.32$ and 0.22, respectively).

Substantively, findings from this analysis indicate that caregivers who are older ($r_s = 0.31$), with a higher income ($r_s = 0.32$), and older child ($r_s = 0.31$) were associated with less objective ($r_s = -0.99$), subjective internalized ($r_s = -0.58$), and subjective
externalized strain \( (r_s = -0.88) \). Caregivers of children in the Sensory Adaptive \( (r_s = 0.57) \) and Taste/Smell Sensitive \( (r_s = 0.47) \) subtypes, when compared with caregivers of children in the Generalized Sensory Difference subtype, were associated with less strain when variance attributable to caregiver age, child age, and household income was held constant. There was not a significant difference between the level of strain experienced by caregivers in the Postural Inattentive and Generalized Sensory Difference subtypes \( (r_s = 0.06) \).
Table 2.5 Standardized coefficients, structural correlations, and squared structural correlations for the first canonical function between caregiver strain and person characteristics

<table>
<thead>
<tr>
<th></th>
<th>Coefficient</th>
<th>$r_s$</th>
<th>$r_s^2$ (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Caregiver strain</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Objective</td>
<td>-1.41</td>
<td>-0.99*</td>
<td>98.01</td>
</tr>
<tr>
<td>Subjective internalized</td>
<td>0.08</td>
<td>-0.58*</td>
<td>33.64</td>
</tr>
<tr>
<td>Subjective externalized</td>
<td>0.39</td>
<td>-0.88*</td>
<td>77.44</td>
</tr>
<tr>
<td><strong>Person characteristics</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Child subtype</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>SA</td>
<td>0.76</td>
<td>0.57*</td>
<td>32.49</td>
</tr>
<tr>
<td>TSS</td>
<td>0.79</td>
<td>0.47*</td>
<td>22.09</td>
</tr>
<tr>
<td>PI</td>
<td>0.47</td>
<td>0.06</td>
<td>0.36</td>
</tr>
<tr>
<td>Caregiver age</td>
<td>0.10</td>
<td>0.31*</td>
<td>9.61</td>
</tr>
<tr>
<td>Child age</td>
<td>0.26</td>
<td>0.31*</td>
<td>9.61</td>
</tr>
<tr>
<td>Household income</td>
<td>0.10</td>
<td>0.32*</td>
<td>10.24</td>
</tr>
<tr>
<td>Number of children</td>
<td>-0.14</td>
<td>-0.16</td>
<td>2.56</td>
</tr>
</tbody>
</table>

Note. Structural correlations ≥ 0.30 are marked with an asterisk (*). $r_s = \text{structural correlation}$; $r_s^2 = \text{squared structural correlation}$; SA = Sensory Adaptive; TSS = Taste/Smell Sensitive; PI = Postural Inattentive; reference group: Generalized Sensory Difference
2.5 Discussion

The objective of the present study was to determine the relationship between child sensory subtype and caregiver burden. Overall, results indicate that caregiver HRQOL does not but caregiver strain does vary as a function of child sensory subtype. In the present study, caregivers of children in different subtypes did not significantly differ in physical and mental HRQOL, as measured by the SF-12v2, when controlling for demographic variables. Findings from studies comparing SF-12v2 scores of caregivers of children with and without ASD, however, have consistently demonstrated that caregivers of children with ASD have poorer mental and physical health than caregivers of typically developing children and children with other developmental disabilities (Khanna et al., 2011; Allik, Larsson, & Smedje, 2006; Eapen & Guan, 2016). These findings suggest that children with ASD in all four sensory subtypes would benefit from routine screening for level of caregiver burden and proactive measures to improve HRQOL.

In the present study, caregivers of children in different subtypes were found to differ in the level of reported objective, subjective internalized, and subjective externalized strain, while controlling for caregiver age, child age, and household income. Specifically, as we hypothesized, caregivers of children in the SA subtype were associated with the lowest levels of strain ($r_s = 0.57$), and caregivers of children in the TSS subtype were associated with an intermediate level of strain ($r_s = 0.47$), when compared to caregivers of children in the GSD subtype. Contrary to our hypothesis, however, caregivers of children in the PI and GSD subtypes were not found to significantly differ in level of caregiver strain. One possible explanation for this finding is that, while the overall analysis was sufficiently powered, we may have been
underpowered for the comparison of the PI (n=77) and GSD (n=133) subtypes, as it is not possible to perform group-specific power analyses. Additionally, this result may be attributable to the theoretical constructs that underlie the four sensory subtypes.

Preliminary findings from our lab support the hypothesis that two key constructs underlie the four sensory subtypes: 1) sensory reactivity, which is the intensity of a response to a stimulus, and 2) multisensory integration, which is the ability to interpret and respond to multiple sensory stimuli simultaneously (Hand, Dennis, Lane, unpublished manuscript; Lane, Eldridge, Hand, Harpster, & Dennis, 2015; Schaaf & Lane, 2015b). Children in the SA subtype do not demonstrate difficulties with sensory reactivity or multisensory integration. Children in the TSS and PI subtypes demonstrate difficulty with sensory reactivity and multisensory integration, respectively, while children in the GSD subtype demonstrate behaviors consistent with both sensory reactivity and multisensory integration deficits. Findings of the present study, in light of this proposed theoretical structure, may suggest that while deficits in either of these areas are associated with increased caregiver strain, difficulties in multisensory integration are associated with a higher level of caregiver strain than are deficits in sensory reactivity.

Although the present study is associational in nature, and thus does not allow for causal inference, findings from qualitative studies of caregivers of children with ASD offer insight into the possible mechanism by which sensory features influence caregiver strain (Bagby, Dickie, & Baranek, 2012; Schaaf et al., 2011). Specifically, these qualitative studies suggest that the child’s sensory features are related to many aspects of family life, including three areas that are captured by items in the objective strain section of the CGSQ: 1) disruption of family routines; 2) disruption of family social activities;
and 3) decreased time spent with other family members. For example, caregivers report having to take two cars on family outings, such as sporting events, as the child with ASD often needs to leave the event before the rest of the family due to an inability to cope with the multisensory nature of the experience (Schaaf et al., 2011). When the child’s sensory features disrupt family outings, caregivers report increased feelings of incompetence, frustration, disappointment, and worry, which also has implications for subjective internalized and externalized strain (Bagby et al., 2012). In addition, caregivers describe avoidance of social situations that will be multisensory in nature (i.e., bowling alleys, outdoor weddings) or in unfamiliar spaces (i.e., another family’s home) as a result of the sensory processing needs of the child with ASD (Bagby et al., 2012; Schaaf et al., 2011). For example, one caregiver reported that her son’s behaviors in response to sensory stimuli made the family “not [want to] attend as many social functions as [they] normally would have” (Bagby et al., 2012, pg. 82). As a lack of perceived social support is known to be predictive of increased levels of burden, these findings may suggest a feed-forward mechanism, whereby sensory features of the child with ASD reduce socialization, which in turn increases caregiver burden (Eapen & Guan, 2016). Moreover, caregivers report that they are able to spend less time with their other child(ren) because the sensory processing needs of the child with ASD require increased attention from one or both parents (Schaaf et al., 2011). Caregivers describe increased feelings of guilt about this disparity in time and attention spent with their typically developing child(ren), which may also influence subjective internalized and externalized strain. These findings from qualitative literature, which directly link sensory processing difficulties of the child to influences on aspects of daily life that are measured by the objective strain section of the
CGSQ, may explain why objective strain had the highest effect size \( r_s^2 = 0.98 \) of the caregiver strain variables.

Moreover, findings from the qualitative literature suggest that one of the biggest barriers to completion of daily routines and occupations is the multisensory aspect of the environment or activity, which may explain why caregivers of children in the two subtypes with difficulties in multisensory integration report similar levels of strain (Bagby et al., 2012; Schaaf et al., 2011). Caregivers describe increased feelings of stress and fear of child elopement or challenging behaviors when taking the child with ASD into multisensory environments in the community, such as a department store (Schaaf et al., 2011). For example, one caregiver reported, “there are things that you say to yourself like this is too big, this room, there are too many people here, it’s too loud, we gotta go” (Schaaf et al., 2011 pg. 11). Caregivers describe the influence of their child’s difficulties with managing multisensory environments on a wide array of daily activities, including: social activities; family leisure outings; family events (i.e., weddings); travel; and shopping. This may explain why caregivers of children in the PI and GSD subtypes, which both demonstrate difficulties with multisensory integration although they are distinct subtypes, did not significantly differ in the level of caregiver strain (Bagby et al., 2012; Schaaf et al., 2011). In contrast, for children in the TSS subtype, the influence of the child’s sensory processing difficulties on the completion of daily routines may be centered predominantly around mealtime. This restricted scope of sensory processing difficulties may explain why caregivers of children in the TSS subtype were found to report significantly less caregiver strain than caregivers of children in the GSD subtype.
2.5.1 Limitations and future directions

Findings of the present study should be considered in light of some limitations; first, the present study relied on caregiver report of ASD diagnosis. For participants recruited through the IAN, we can be confident that the child did receive an independent ASD diagnosis due to the recent validation of diagnoses in the registry. However, there is less certainty with regard to participants who responded to waiting room fliers and ResearchMatch emails. Therefore, future replication of this study in a sample of children with independently confirmed ASD diagnoses is advisable.

Additionally, the number of participants belonging to the SA subtype was disproportionately small in the present sample when compared with other independent samples of children with ASD (Lane et al., 2014a). We hypothesize that this could be due to two primary reasons. First, there may have been sampling bias as a result of recruitment materials that mentioned an interest in “sensory processing” and caregiver burden. While the materials did state that we were looking for caregivers of children with ASD with and without sensory difficulties, mention of this special interest may have decreased the likelihood of response by caregivers of children in the SA subtype. Second, the use of clinical recruitment sites may have been another source of sampling bias, as it is possible that children in the SA subtype may not receive outpatient occupational, physical, or speech therapy services. Thus, caregivers of children in the SA subtype may not have been presented with the same opportunities for participation as caregivers of children in other subtypes. Future studies seeking to replicate these findings should carefully consider these aspects of recruitment in seeking to recruit more caregivers of children in the SA subtype.
Finally, there are some limitations associated with the statistical analysis used in the present study. As CCA is associational in nature, we are unable to assert any causal relationships between sensory subtypes and level of caregiver burden. We can, however, conclude that caregivers of children in different subtypes are associated with different levels of perceived strain. Moreover, findings from qualitative literature provide valuable insight as to how sensory processing and caregiver strain may be related. Additionally, it is noteworthy that CCA reflects the variance shared by the canonical variates, which are the linear composites of observed variables, rather than the observed variables themselves. Therefore, it is recommended that the present study be replicated with a larger sample size that would sufficiently power alternate statistical methodologies (i.e., MANCOVA). Lastly, there may be some concern that the present study was overpowered, as our power analysis indicated that a sample size of 282 would be sufficient to detect a small effect at 80% power. In order to test the possible effect of a large sample size, the CCA was re-run with a random selection of 75% (n = 275) of the data. This analysis revealed that the substantive interpretation remained the same, which lends credibility to the robustness of these findings.

2.6 Summary and Conclusion

The present study did not detect an association between sensory subtypes and level of caregiver perceived mental and physical health. This may suggest that caregivers of children in all subtypes would benefit from interventions targeting these areas, as it is known that caregivers of children with ASD have poorer physical and mental HRQOL than caregivers of typically developing children and children with other developmental disabilities. Level of caregiver objective, subjective internalized, and subjective
externalized strain were associated with the child’s sensory subtype, while controlling for caregiver age, child age, and household income. When compared with caregivers of children in the GSD subtype, caregivers of children in the SA subtype reported the lowest levels of caregiver strain, followed by caregivers of children in the TSS subtype. Caregivers of children in the PI and GSD subtypes did not significantly differ in level of caregiver strain. Increasing caregiver and child age, as well as higher household income, were associated with less caregiver strain.

Findings of the present study may assist in alleviating burden of caregivers of children with ASD by alerting clinicians to aspects of caregiver burden associated with the child’s sensory subtype to target for intervention. These findings, when combined with existing literature, have the capability to support decision making for clinicians working with children with ASD and their families by linking each subtype with intervention targets for the child and caregiver. Ultimately, this may lead to improved treatment outcomes for children with ASD and their families.
3.1 Abstract

Background: Difficulties interpreting and responding to everyday environmental sensory stimuli are known to influence the participation of children with autism spectrum disorder (ASD) in everyday meaningful activities. Recently, researchers have suggested that children with ASD can be classified into one of four sensory subtypes based on shared patterns of sensory difficulties: Sensory Adaptive; Taste/Smell Sensitive; Postural Inattentive; and Generalized Sensory Difference. However, the relationship of a child’s sensory subtype and level of participation remain unexplored. Purpose: The purpose of the present study is to examine the relationship between sensory subtypes of children with ASD and the child’s participation in home, school, and community activities. Method: A national online survey of caregivers of children with ASD was conducted (n = 369). Canonical correlation analysis was used to examine the relationship between sensory subtypes and indicators of child participation including frequency and level of involvement in home, school, and community activities. Results: The analysis revealed that child participation in home, school, and community activities was significantly associated with the child’s sensory subtype membership such that children in the Sensory Adaptive subtype were associated with the highest levels of participation, followed by children in the Taste/Smell Sensitive subtype, when compared with children in the
Generalized Sensory Difference subtype. Children in the Postural Inattentive and Generalized Sensory Difference subtypes did not significantly differ in levels of participation. **Conclusions:** This study demonstrates that participation patterns in ASD are associated with sensory subtypes. Possible mechanisms for the relationship between participation and sensory subtype membership are discussed.
3.2 Introduction

According to the International Classification of Functioning, Disability, and Health (ICF), participation refers to one’s involvement in life situations (World Health Organization, 2001). In addition to being a key health-related domain in the ICF, participation in daily activities in the home, school, workplace, and community is an essential ingredient of occupational therapy evaluation and intervention. The Occupational Therapy Practice Framework defines participation as active involvement in daily life activities that the individual finds purposeful and meaningful (AOTA, 2014). Participation is a complex multidimensional construct that can be captured by objective indicators, such as frequency of participation in a particular activity or type of activity (Coster et al., 2012; McConachie, Colver, Forsyth, Jarvis, & Parkinson, 2006). Additionally, it is important to consider more qualitative indicators, such as level of involvement (i.e., attention, nonverbal behavior, being engaged) in an activity, satisfaction with participation, or desire for change (Bedell, Khetani, Cousins, Coster, & Law, 2011; Brown et al., 2004; Coster et al., 2012). Participating in valued activities affords children opportunities for learning and developing skills and improves quality of life (Dunst, Bruder, Trivette, & Hamby, 2006; Humphrey & Wakeford, 2006; The Whoqol Group, 1998). Moreover, increased participation has been associated with reduced incidence of behavioral and emotional difficulties, improved engagement in school, and improved social relationships (Law, 2002).

Children with autism spectrum disorder (ASD) are known to participate less frequently in activities with less variety compared to typically developing children and children with other developmental disabilities (LaVesser, & Berg, 2011; Potvin, Snider,
Prelock, Kehayia, & Wood-Dauphinee, 2012). Moreover, children with ASD demonstrate significantly less competence than typically developing peers in performing the activities in which they do participate (Reynolds et al., 2011). Individual characteristics, including language ability, autism severity, and cognitive function, are associated with the level of participation demonstrated by children with ASD (Little et al., 2015; Orsmond et al., 2013; Rosenberg et al., 2012). Sensory features are atypical responses to everyday environmental stimuli and are another child characteristic that is associated with participation (Little et al., 2015; Schaaf & Lane, 2015). Sensory features are prevalent in ASD and may manifest in a variety of ways. A child may: demonstrate hyper- or hypo-reactivity to sensory stimuli; have unusual sensory interests; inaccurately perceive sensory stimuli; and/or have difficulties managing multiple sensory stimuli simultaneously (Schaaf & Lane, 2015). For example, a child with hypo-reactivity to auditory stimuli may not orient when his or her name is called, whereas a child with hyper-reactivity to an auditory stimulus may cover their ears to dampen sounds that are not perceived to be loud by others. A child who has difficulties with visual perception may not accurately perceive facial expressions and body language that convey emotion, while a child with unusual interest in visual stimuli may hold objects up to the corners of their eyes, or view objects through squinted eyelids. Lastly, a child with difficulties in multisensory integration may have difficulty concentrating in busy or loud environments due to an abundance of auditory, visual, and perhaps tactile or olfactory stimuli.

Findings from existing literature support the notion that sensory features differentially influence participation in valued activities in children with and without ASD. Bar-Shalita and colleagues (2008) found that children with sensory modulation
disorder, without ASD diagnoses, had less frequent activity performance and less activity enjoyment than children without sensory processing difficulties. Literature on the relationship between sensory features and participation in ASD has focused on the type of sensory processing difficulty that the child experiences. Little and colleagues (2015) reported that children with ASD and enhanced sensory perception participate more frequently in activities across home, community, and outdoor contexts, while hyper-reactivity has a significant negative effect on activity participation. Further, these authors concluded that children with increased hypo-reactivity are more likely to participate in activities outside of the home, while those with increased sensory interests, repetitions, and seeking behaviors are more likely to participate in activities inside the home (Little et al., 2015).

Recently, researchers have reported that individuals with ASD can be classified into distinct sensory subtypes based on shared patterns of sensory features (Ausderau et al., 2014; Lane et al., 2010, 2011, 2014a). There are multiple potential advantages to the use of sensory subtypes in understanding functional limitations in ASD, as this methodology groups similar individuals rather than similar behaviors. Subsequently, sensory subtypes offer the potential for tailored interventions based in individual profiles (The Interagency Autism Coordinating Committee, 2014). The most widely researched sensory subtypes are those described by Lane and colleagues (Lane et al., 2010, 2011, 2014a). Lane et al. (2014a) propose four sensory subtypes based on parent responses to the Short Sensory Profile (SSP), a 38-item parent questionnaire (McIntosh, Miller, Shyu, & Dunn, 1999). The subtypes are: 1) Sensory Adaptive; 2) Taste/Smell Sensitive; 3) Postural Inattentive; and 4) Generalized Sensory Difference (Lane et al., 2014a). The
sensory subtypes differ on the severity and focus of sensory features. Severity can be best mapped to performance on the auditory filtering and under-responsive/seeks sensation sensory domains; children in the Sensory Adaptive (SA) subtype are characterized as mild, Taste/Smell Sensitive (TSS) and Postural Inattentive (PI) are characterized as moderate, and Generalized Sensory Difference (GSD) are characterized by severe difficulty in these domains. The two subtypes with moderate severity in the auditory filtering and under-responsive/seeks sensation SSP domains, TSS and PI, are further differentiated by their focus. Specifically, children in the TSS subtype demonstrate an extreme score in the taste/smell sensitivity domain, while children in the PI subtype display difficulties in maintaining postural integrity, as evidenced by extreme scores in the SSP domain of low energy/weak.

Additionally, distinctive non-sensory behavior profiles are associated with each sensory subtype. Children in the TSS subtype, for example, are pickier eaters and have more severe communication impairments, while children in the GSD subtype demonstrate more maladaptive behaviors (i.e., sucking on one’s thumb or fingers, blaming mistakes on others) (Lane et al., 2010, 2011). The subtypes have also been documented to differ in daily living skills, emotional regulation, and attention (Tanner & Lane, unpublished manuscript). Despite these distinct behavior profiles, systematic variation between subtypes in age, non-verbal IQ, or severity of autistic symptoms, including social interaction, communication, restricted and repetitive behaviors, has not been found (Lane et al., 2014a). Associating sensory subtypes with functional performance limitations may guide clinical decision-making, which may subsequently improve intervention effectiveness. One area of functional performance that has not yet
been explored as a function of sensory subtype is participation in home, school and community activities. Given that children in different sensory subtypes vary on several personal characteristics and behaviors, it is likely that they differ in participation in valued activities. Additional information about participation as a function of sensory subtype may be useful for clinicians working with children with ASD by identifying potential domains of participation in need of intervention for each sensory subtype.

As a next step toward the widespread clinical use of sensory subtyping to guide intervention selection for children with ASD, the objective of the present study is to examine participation in home, school, and community activities as a function of sensory subtype. Specifically, we will determine differences between subtypes on frequency of participation and level of involvement in home, school, and community activities. Our hypothesis is that children in subtypes with mild sensory features will have higher frequency of participation and level of involvement in home, school, and community activities than those with more severe sensory features. In other words, we hypothesize that children in the SA subtype will demonstrate the highest levels of participation, children in the TSS and PI subtypes will demonstrate intermediate levels of participation, and children in the GSD subtype will have the lowest levels of participation.

3.3 Materials and Methods

3.3.1 Study Design

A non-experimental, cross-sectional design was used to achieve our objective. The primary variables of interest were: 1) participation in home, school, and community activities, which includes frequency and intensity of participation, as measured by the Participation and Environment Measure- Children and Youth, and 2) sensory subtype of
the child for whom the participant provides care, as measured by the Short Sensory Profile. The child’s age, household income, and number of children in the household were included as covariates due to the association of these variables with participation in other populations (Dunn, Coster, Cohn, & Ormond, 2009; King et al., 2006; Rosenberg et al., 2012).

3.3.1 Participants

Participants were primary caregivers of children aged 5-13 years with ASD (n = 369). A primary caregiver was defined as the person who takes primary responsibility for the child with ASD. To eliminate potential confounding effects on child participation, participants were excluded if their child had a comorbid neurological diagnosis (i.e., cerebral palsy, stroke, spinal cord injury), genetic diagnosis (i.e., Rett, Down, or Fragile X syndromes), and/or a significant physical disability, as these conditions could also account for participation limitations.

Participants for the present study were recruited through the Interactive Autism Network (IAN), ResearchMatch, and pediatric clinics in the greater Columbus, Ohio area. The IAN is an online research registry for caregivers of children with ASD that recently reviewed medical records for a subset of children in the registry to authenticate parent-report ASD diagnoses (Daniels et al., 2011). Based on the inclusion criteria, a total of 8,215 registrants in the IAN were approached for participation in the study. ResearchMatch is a national online registry for individuals interested in participating in research to be matched with ongoing research studies. Based on inclusion criteria, 255 participants in ResearchMatch were contacted for participation. Participants meeting study criteria from the IAN and ResearchMatch received a brief study description,
including a link to the survey site via email. To maximize study response rate, a total of three emails were sent to each potential participant. The present study was also advertised in pediatric occupational, physical, and speech therapy clinics by posting fliers in waiting rooms and reception areas. Figure 3.1 provides a flow chart illustration of the survey response rate and retention process during survey completion. Participants who completed the survey received a Redbox DVD code via email. The Institutional Review Board of The Ohio State University reviewed and approved this study.
Figure 3.1 Child participation survey response rate

- Individuals who received emails, n=8,470
- Individuals who completed informed consent, n=631
- Completed inclusion criteria screener, n=593
  - Participants excluded:
    - Neurologic comorbidity (n=115)
    - Genetic comorbidity (n=52)
    - Physical disability (n=15)
  - Attrition (n=34)
- Completed SSP, n=378
- Completed PEM-CY, n=378
- Completed demographics, n=378

*Note. SSP = Short Sensory Profile; PEM-CY = Participation and Environment Measure – Children and Youth*
3.3.2 Measures

*Sensory Subtype Classification.*

Caregivers completed the Short Sensory Profile for the purpose of determining child sensory subtype (McIntosh et al., 1999). The SSP consists of 38 items that are designed to measure behaviors associated with abnormal sensory processing in seven sensory domains: 1) tactile; 2) taste/smell; 3) movement; 4) visual/auditory sensitivity; 5) under-responsive/seeks sensation; 6) auditory filtering; and 7) low energy/weak. Scores for the seven sensory domains of the SSP are compared to normative data from 1,200 typically developing children. Higher domain scores indicate more typical sensory processing and lower scores indicate a probable or definite difference in sensory processing. The SSP demonstrates moderate to excellent internal consistency of overall and subdomain sections ($r = 0.70 – 0.90$) and acceptable discriminative validity ($> 95\%$ in differentiating children with and without sensory impairments, McIntosh et al., 1999). While the SSP was initially designed and validated for measuring sensory processing in children aged 3-10 years, it has been widely used in research for children over 10 years of age (Mangeot et al., 2001; Schoen et al., 2008; Tavassoli et al., 2016; Uljarević et al., 2016). Additionally, the SSP demonstrates convergent validity with conceptually similar measures in individuals with ASD up to 14 years of age (Tavassoli et al., 2016), and in individuals with sensory processing difficulties, but not ASD, up to 16 years of age (Schoen et al., 2008). As a result, a broader age range of participants is reflected in the present sample.
Participation.

Participation in home, school, and community activities was measured with the Participation and Environment Measure- Children and Youth (PEM-CY, Coster et al., 2014). The PEM-CY is a 70-item parent report measure of children’s level of participation in home, school, and community activities. It is intended for administration to parents of children aged 5-17 years and measures both frequency of participation (i.e., how often a child participates) and level of involvement (i.e., attention, engagement). Internal consistency reliability has been demonstrated to be adequate (reliability coefficients range from 0.59-0.83) and test-retest reliability was moderate to good (≥0.58). Construct validity was established using the known groups method, where groups of individuals who are hypothesized to differ in levels of participation are found to differ in PEM-CY scores (Coster et al., 2011; Coster et al., 2014).

3.3.3 Data analyses

Participants with over 15% missing data (n = 3) or who did not report child age (n = 3) were excluded from analyses. Participants missing 50% or more data from any single domain of the SSP (n = 3) were also excluded from the analysis due to potential impact on subtype determination. Missing values for remaining participants were imputed with iterative model-based imputation using robust stepwise regression with the package “VIM” in the statistical software R (Templ et al., 2015). As an additional validation procedure, because the effect of imputing missing SSP values on sensory subtype determination has not yet been tested, a content area expert on sensory subtypes examined SSP domain z-scores for participants with missing values (n = 25) prior to imputation. Based on visual inspection of z-scores, the expert recorded the subtype in
which they hypothesized each participant belonged. There was a 68% agreement rate between the expert’s visual inspection and the algorithm-identified subtype classification based on the imputed values. While this was a relatively conservative level of agreement, the participants with missing values were included in subsequent analyses because the missing data rate for the SSP was very small (0.24%).

Participant-level scores for mean frequency and level of involvement in home, school, and community activities were calculated following the directions in the PEM-CY Manual (Coster et al., 2014). Participants were classified into sensory subtypes using the algorithm published by Lane and colleagues based on domain level z-scores.

Demographic information was analyzed descriptively. Lastly, canonical correlation analysis (CCA), a multivariate generalized linear modeling technique, was conducted to explore the relationship between participation and person characteristics.

Canonical correlation analysis can be conceptualized similarly to multiple regression (Hair, 2006). CCA, however, allows for the simultaneous examination of several dependent variables and several independent variables by producing a linear combination of each set of variables (independent and dependent) that maximizes the correlation between the variable sets (Hair, 2006; Dardas & Ahmad, 2014). Similar to multiple regression, the variance for each variable that is attributable to the other variables in the set is controlled. The linear combinations of the observed variables, called canonical functions, can be interpreted like multiple regression equations (Sherry & Henson, 2005). CCA generates an equal number of canonical functions to the number of variables in the smaller of the two variable sets. Each canonical function yields a measure of the relationship between the independent and dependent variables, called a
canonical correlation. A canonical correlation can be conceptualized as a simple Pearson correlation (Sherry & Henson, 2005).

There are multiple benefits to the use of CCA as opposed to univariate statistical analyses. First, because CCA is a multivariate technique, it limits the probability of Type I error (Fish, 1988). Additionally, as child participation is a complex construct that is influenced by multiple variables and the interaction of those variables, this technique allows for the detection of important multivariate relationships that may not be captured when using univariate methods.

In the present study, CCA was performed to examine the relationship between a set of participation variables (frequency and level of involvement in home, school, and community activities) and a set of personal characteristics (sensory subtype, age, household income, and number of children in the household). As sensory subtype is a categorical variable, a dummy coding scheme, which uses ones and zeros to convey information about group membership, was used with GSD as the reference group. Dummy coding creates \( k-1 \) new variables, where \( k \) = the number of groups, to represent a categorical variable. In this case, three dummy variables were created. Participants belonging to the GSD subtype were assigned a score of zero on each of the dummy variables. Participants in the SA, TSS, and PI subtypes were each assigned a score of one to their respective dummy variables and zero on the remaining dummy variables. Figure 3.2 provides a graphic illustration of the CCA conducted in the present study. All analyses were conducted using the statistical software program R with ad-on package “yacca” (R Development Core Team, 2011; Butts & Butts, 2009).
Figure 3.2 Schematic representation of canonical correlation analysis for child participation and personal characteristics

Note. SA = Sensory Adaptive; TSS = Taste/Smell Sensitive; PI = Postural Inattentive; GSD = Generalized Sensory Difference (reference group)
Results of the CCA were interpreted using a multi-step procedure by first evaluating the overall significance of the full model via significance tests of Wilk’s $\lambda$ (Sherry & Henson, 2005). Next, the statistical and practical significance of individual functions was evaluated with Wilk’s $\lambda$ and squared canonical correlations ($R_c^2$), which are variance-accounted for effect sizes and are analogous to $R^2$ effects in multiple regression. Last, the importance of individual variables were interpreted for significant functions using the following values: 1) standardized canonical coefficients, which are the coefficients from the linear equations that combine the observed independent and dependent variables into respective synthetic variables; 2) structural correlations, which are bivariate correlations between the observed and synthetic variables; and 3) squared structural correlations, which are the amount of variance an observed variable linearly shares with its respective synthetic variable. A value of $\geq 0.30$ for structural correlations indicated significance (Dardas & Ahmad, 2014; Matson, Neal, Fodstad, & Hess, 2010).

*Power analysis.*

An a-priori power analysis for a Pearson’s $r$ was conducted using G*Power 3.1 (Faul et al., 2009). The sample size estimate to capture a small effect (Cohen’s $q = 0.30$) with 80% power, is $n = 282$ (Dardas & Ahmad, 2014).

### 3.4 Results

A total of 369 usable survey responses were received. Table 3.1 provides descriptive information about the participants. The average age of children included in the study was 10.10 years (SD = 2.52) and the majority (93.77%) were male. Caregivers completing the surveys were, on average, 42.49 years of age (SD = 6.89), and 83.20% were female. A majority of participants were married (82.11%), 10.03% were divorced,
and 7.31% were single. Nearly 97% had either attended or graduated from college and 69% were employed full or part time. Descriptive statistics for participant scores on the PEM-CY for all participants and by subtype are detailed in Table 3.2. Bivariate correlations for the person characteristic variables and participation variables are shown in Table 3.3.
<table>
<thead>
<tr>
<th></th>
<th>All</th>
<th>SA</th>
<th>TSS</th>
<th>PI</th>
<th>GSD</th>
</tr>
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<tbody>
<tr>
<td>n</td>
<td>369</td>
<td>20</td>
<td>137</td>
<td>78</td>
<td>134</td>
</tr>
<tr>
<td>Mean age (SD)</td>
<td>10.10 (2.52)</td>
<td>10.19 (2.57)</td>
<td>9.95 (2.67)</td>
<td>10.25 (2.52)</td>
<td>10.13 (2.36)</td>
</tr>
<tr>
<td>Age range</td>
<td>4.95-13.98</td>
<td>5.29-13.76</td>
<td>5.01-13.95</td>
<td>5.35-13.98</td>
<td>4.95-13.88</td>
</tr>
<tr>
<td>% Male</td>
<td>93.77</td>
<td>95.00</td>
<td>90.51</td>
<td>96.15</td>
<td>95.52</td>
</tr>
<tr>
<td>Median number of children (SD)</td>
<td>2 (0.99)</td>
<td>2 (0.75)</td>
<td>2 (1.10)</td>
<td>2 (0.97)</td>
<td>2 (0.92)</td>
</tr>
<tr>
<td>Household income*</td>
<td>100k+</td>
<td>100k+</td>
<td>100k+</td>
<td>100k+</td>
<td>25-50k</td>
</tr>
<tr>
<td>Caregiver mean age (SD)</td>
<td>42.49 (6.89)</td>
<td>43.68 (3.66)</td>
<td>42.95 (7.12)</td>
<td>41.80 (6.39)</td>
<td>42.24 (7.30)</td>
</tr>
<tr>
<td>Caregiver age range</td>
<td>25.82-66.80</td>
<td>36.20-50.02</td>
<td>28.74-66.80</td>
<td>25.82-61.28</td>
<td>29.08-61.18</td>
</tr>
</tbody>
</table>

*Mode. SA = Sensory Adaptive; TSS = Taste/Smell Sensitive; PI = Postural Inattentive; GSD = Generalized Sensory Difference; SD = Standard deviation
Table 3.2: Descriptions of PEM-CY scores

<table>
<thead>
<tr>
<th></th>
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<th>SA</th>
<th>TSS</th>
<th>PI</th>
<th>GSD</th>
</tr>
</thead>
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<tr>
<td><strong>Frequency of participation (Scoring Range 0 - 7)</strong></td>
<td></td>
<td></td>
<td></td>
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</tr>
<tr>
<td><strong>Home</strong></td>
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<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mean</td>
<td>5.25</td>
<td>5.53</td>
<td>5.34</td>
<td>5.29</td>
<td>5.09</td>
</tr>
<tr>
<td>Median</td>
<td>5.33</td>
<td>5.28</td>
<td>5.40</td>
<td>5.35</td>
<td>5.20</td>
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<td>2.43-7.00</td>
<td>3.50-7.00</td>
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</tr>
<tr>
<td>IQR</td>
<td>4.89-5.70</td>
<td>5.28-5.93</td>
<td>5.00-5.67</td>
<td>5.00-5.71</td>
<td>4.60-5.69</td>
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<td><strong>School</strong></td>
<td></td>
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<td></td>
<td></td>
</tr>
<tr>
<td>Mean</td>
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<td>4.36</td>
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<td>3.75-5.25</td>
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<tr>
<td>Mean</td>
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<td>4.12</td>
<td>3.75</td>
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<td>3.38</td>
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<td>3.20-3.90</td>
<td>2.93-3.98</td>
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<td><strong>School</strong></td>
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<tr>
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<td>3.00</td>
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<td>0.00-5.00</td>
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<td>IQR</td>
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<td>2.50-3.75</td>
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<td><strong>Community</strong></td>
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<td></td>
<td></td>
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<tr>
<td>IQR</td>
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<td>3.38-4.40</td>
<td>2.67-3.67</td>
<td>2.50-3.83</td>
<td>2.25-3.32</td>
</tr>
</tbody>
</table>

Note. SA = Sensory Adaptive; TSS = Taste/Smell Sensitive; PI = Postural Inattentive; GSD = Generalized Sensory Difference; IQR = Interquartile range
Table 3.3 Bivariate correlations between participation and person characteristics

<table>
<thead>
<tr>
<th></th>
<th>Frequency</th>
<th>Involvement</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Home</td>
<td>School</td>
</tr>
<tr>
<td>SA (vs. GSD)</td>
<td>0.09</td>
<td>0.09</td>
</tr>
<tr>
<td>TSS (vs. GSD)</td>
<td>0.09</td>
<td>0.04</td>
</tr>
<tr>
<td>PI (vs. GSD)</td>
<td>0.03</td>
<td>0.03</td>
</tr>
<tr>
<td>Age</td>
<td>-0.11*</td>
<td>0.12*</td>
</tr>
<tr>
<td>Household income</td>
<td>0.02</td>
<td>0.17†</td>
</tr>
<tr>
<td>Number of children</td>
<td>0.08</td>
<td>0.01</td>
</tr>
</tbody>
</table>

Note. *p < 0.05; †p ≤ 0.01; SA = Sensory Adaptive; TSS = Taste/Smell Sensitive; PI = Postural Inattentive; GSD = Generalized Sensory Difference
The assumption of normality for CCA was examined visually and descriptively. All variables, with the exception of number of children in the household, were reasonably normally distributed. Number of children in the household was found to be positively skewed; however, as the impact of non-normality on multivariate analyses effectively diminishes with sample sizes of at least 200, the present analysis is suitable for interpretation (Hair, 2006, p. 86). While not a requirement for CCA, the absence of multicollinearity was evaluated (Nimon et al., 2010). Examination of the bivariate correlations within and between the person and participation variable sets revealed an absence of multicollinearity (within-set correlations $r \leq 0.54$; between-set correlations $r \leq 0.21$).

Collectively, the full model was statistically significant (Wilk’s $\lambda = 0.80$, $\chi^2(36) = 84.56$, $p < 0.0001$). Evaluation of the $R^2_c$ revealed that the full model explained 20% of the variance shared between the two variable sets ($R^2_c = 0.20$). Function 1 accounted for over 10% of the variance explained ($R^2_c = 0.103$). While Function 2, when tested in isolation, was statistically significant (Wilk’s $\lambda = 0.89$, $\chi^2(25) = 41.10$, $p < 0.05$), it accounted for less than 6% of the variance explained ($R^2_c = 0.052$), and thus was not practically significant. As a result, Function 2 was not considered in subsequent interpretation. The remaining functions, when tested in isolation, were not statistically significant (Table 3.4).
Table 3.4 Results of the six canonical correlations

<table>
<thead>
<tr>
<th>Function</th>
<th>Wilk’s $\lambda$</th>
<th>$\chi^2$</th>
<th>df</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>0.80</td>
<td>84.56†</td>
<td>36</td>
</tr>
<tr>
<td>2</td>
<td>0.89</td>
<td>41.10*</td>
<td>25</td>
</tr>
<tr>
<td>3</td>
<td>0.94</td>
<td>21.75</td>
<td>16</td>
</tr>
<tr>
<td>4</td>
<td>0.97</td>
<td>10.59</td>
<td>9</td>
</tr>
<tr>
<td>5</td>
<td>0.99</td>
<td>4.15</td>
<td>4</td>
</tr>
<tr>
<td>6</td>
<td>1.00</td>
<td>0.55</td>
<td>1</td>
</tr>
</tbody>
</table>

*Note.* *p* < 0.05; †*p* < 0.0001; $\chi^2$ = Chi-square statistic; df = degrees of freedom
Results for the first canonical function (Table 3.5) indicate that all participation variables had significant structural correlations, however the effect size was highest for community involvement \((r_s^2 = 0.72)\). The significant variables from the person characteristics set were SA, TSS, and household income. The SA variable had the largest effect size of the person characteristic variables \((r_s^2 = 0.50)\). Substantively, these results indicate that children with ASD who belong to the SA \((r_s = 0.71)\) and TSS \((r_s = 0.36)\) subtypes, when compared with the GSD subtype, demonstrate significantly more frequent participation in home \((r_s = 0.46)\), school \((r_s = 0.47)\), and community activities \((r_s = 0.52)\), as well as higher levels of involvement in the activities in which they do participate in the home \((r_s = 0.62)\), school \((r_s = 0.44)\), and community \((r_s = 0.85)\), when variance attributable to household income is held constant. No significant difference in participation was detected between members of the PI and GSD subtypes \((r_s = 0.11)\). Children from families with higher incomes \((r_s = 0.31)\) were associated with higher levels of participation across settings. The number of children living in the household and the age of the child with ASD were not significantly associated with frequency of participation and level of involvement in home, school, and community activities.
Table 3.5 Standardized coefficients, structural correlations, and squared structural correlations for the first canonical function between participation and person characteristics

<table>
<thead>
<tr>
<th>Participation</th>
<th>Coefficient</th>
<th>$r_s$</th>
<th>$r_s^2$ (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Home frequency</td>
<td>0.27</td>
<td>0.46*</td>
<td>21.16</td>
</tr>
<tr>
<td>School frequency</td>
<td>0.33</td>
<td>0.47*</td>
<td>22.09</td>
</tr>
<tr>
<td>Community frequency</td>
<td>0.03</td>
<td>0.52*</td>
<td>27.04</td>
</tr>
<tr>
<td>Home involvement</td>
<td>0.28</td>
<td>0.62*</td>
<td>38.44</td>
</tr>
<tr>
<td>School involvement</td>
<td>-0.14</td>
<td>0.44*</td>
<td>19.36</td>
</tr>
<tr>
<td>Community involvement</td>
<td>0.71</td>
<td>0.85*</td>
<td>72.25</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Person characteristics</th>
<th>Coefficient</th>
<th>$r_s$</th>
<th>$r_s^2$ (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>SA</td>
<td>0.89</td>
<td>0.71*</td>
<td>50.41</td>
</tr>
<tr>
<td>TSS</td>
<td>0.71</td>
<td>0.36*</td>
<td>12.96</td>
</tr>
<tr>
<td>PI</td>
<td>0.51</td>
<td>0.11</td>
<td>1.21</td>
</tr>
<tr>
<td>Age</td>
<td>0.08</td>
<td>0.09</td>
<td>0.81</td>
</tr>
<tr>
<td>Household income</td>
<td>0.12</td>
<td>0.31*</td>
<td>9.61</td>
</tr>
<tr>
<td>Number of children</td>
<td>-0.11</td>
<td>-0.08</td>
<td>0.64</td>
</tr>
</tbody>
</table>

*Note. Structural correlations ≥ 0.30 are marked with an asterisk (*). $r_s$ = structural correlation; $r_s^2$ = squared structural correlation; SA = Sensory Adaptive; TSS = Taste/Smell Sensitive; PI = Postural Inattentive; reference group: Generalized Sensory Difference (GSD)*
3.5 Discussion

The purpose of this study was to examine whether participation in home, school and community activities varies by sensory subtype. Results suggest that children in different sensory subtypes differ in frequency of participate in home, school, and overall level of involvement in activities. Specifically, the SA subtype was associated with the highest levels of participation ($r_s = 0.71$), followed by the TSS subtype ($r_s = 0.36$), when compared with children in the GSD subtype. There was not a significant difference in levels of participation of children in the PI and GSD subtypes. This finding is surprising, given that examination of PEM-CY score descriptives (Table 3.2) may result in the impression that children in the PI subtype demonstrated participation patterns similar to that of children in the TSS subtype, which were found to differ from the GSD subtype. There are two potential statistical explanations for this result. First, it is possible that, although the overall analysis was sufficiently powered, the direct comparison between the PI (n = 78) and GSD (n = 134) subtypes was underpowered, as there is not currently a way to perform group-specific power analyses. Second, it is possible that the PI and GSD subtypes did not significantly differ due to the structure and nature of the synthetic variables in the CCA. Examination of the coefficients (Table 3.5) of the linear equations used to derive the synthetic variables reveals that the participation variable is comprised predominantly of the variables: home frequency (coef = 0.27), school frequency (coef = 0.33), home involvement (coef = 0.28), and community involvement (coef = 0.71). On two of these observed variables, home frequency and home involvement, the TSS subtype demonstrated slightly higher median scores (5.35 and 3.58, respectively) than children in the PI subtype (5.20 and 3.44, respectively), as described in Table 3.2. This
may explain why the TSS subtype had a higher structural correlation ($r_s = 0.36$) with the participation synthetic variable, and is statistically significantly different from the GSD subtype, while the PI subtype is not ($r_s = 0.11$).

Another possible explanation for this finding is the hypothesized theoretical constructs that underlie the four sensory subtypes. Preliminary work from our lab suggest that two key constructs underlie the sensory subtypes: 1) sensory reactivity, which is the intensity of a response to a stimulus, and 2) multisensory integration, which is the ability to interpret and respond to multiple sensory stimuli simultaneously (Hand, Dennis, & Lane, unpublished manuscript; Lane, Eldridge, Hand, Harpster, & Dennis, 2015; Schaaf & Lane, 2015b). Children in the SA subtype do not demonstrate difficulties with either sensory reactivity or multisensory integration. The TSS subtype is associated with difficulties in sensory reactivity, while the PI subtype is associated with challenges in multisensory integration. Children in the GSD subtype display behaviors consistent with deficits in both sensory reactivity and multisensory integration. In light of this proposed theoretical structure, results of this study may suggest that while deficits in either of these areas are associated with decreased participation in home, school, and community activities, difficulties in multisensory integration are associated with lower levels of participation than are deficits in sensory reactivity.

While the present analysis is associational in nature and does not support causal inference, findings from existing literature may help to elucidate the mechanism by which sensory processing and participation are related. One possible explanation for the finding that children in the subtypes with multisensory integration deficits, PI and GSD, did not significantly differ in levels of participation is the impact that multisensory
integration is thought to have on motor performance (Izawa et al., 2012; Sober & Sabes, 2003; White, Mulligan, Merrill, & Wright, 2007). In a study of children with and without sensory processing difficulties, but without ASD, White and colleagues (2007) suggest that the ability to process and integrate visual, vestibular, and tactile information supports a child’s ability to perform the motor actions necessary to interact with objects in the environment. These motor actions include: bending; coordinating movements; object manipulation; modulation of movement and force; and stabilizing (White et al., 2007). A different study that examined motor learning found that children with ASD demonstrate a weaker association between motor commands and visual feedback when compared to children with attention-deficit hyperactivity disorder and typically developing children, which may contribute to the development of motor dyspraxia in children with ASD (Izawa et al., 2012). It is reasonable to conjecture, therefore, that difficulties with performing coordinated motor actions may reduce a child’s frequency of participation and level of involvement in many activities that require such actions, including household chores, organized sports, and unstructured physical activities (i.e., playing catch, bicycle riding).

In addition to poorer motor coordination, literature suggests that children with ASD have difficulties with the temporal processing of multisensory stimuli, which may be a barrier to participation. Foss-Feig and colleagues (2010), for example, found that children with ASD interpreted auditory and visual stimuli as occurring simultaneously over a wider range of stimulus onset asynchronies when compared to children without ASD. These difficulties in temporal audiovisual processing for children with ASD have been documented to rise with increasing auditory complexity (i.e., speech sounds) and
may negatively affect social interaction, which requires precise interpretation of facial expressions, body language, and vocal tones (Stevenson et al., 2014; Foss-Feig et al., 2010). In regard to participation of children with ASD and multisensory integration deficits, difficulties with timing audiovisual information may result in decreased social activities, including interacting with peers and participating in group work at school, as well as other activities that require precise audio-visual timing, such as playing video games, and taking special classes and lessons (i.e., music, art). Taken together, findings from studies on motor coordination and audio-visual temporal processing support the notion that, while PI and GSD are distinct sensory subtypes, shared limitations in multisensory integration may explain why a significant difference in participation was not detected between these subtypes.

Our findings also suggest that children in the TSS subtype, who do not demonstrate difficulties with multisensory integration, have lower levels of participation than members of the SA subtype, as evidenced by a smaller structural correlation. Existing literature suggests that children with ASD and taste/smell sensitivity dysfunction limit the amount of time spent participating in recreational activities that are associated with intense smell stimuli, such as doing crafts, playing with, and caring for, pets (Hoddenbach et al., 2012). It is reasonable to suggest, therefore, that children in the TSS subtype may avoid participating in types of activities measured by the PEM-CY that are associated with intense smells including arts and crafts (i.e., painting, pottery), performing household chores (i.e., taking out the garbage, cooking, cleaning, caring for pets), neighborhood outings (i.e., going to restaurants or malls), and organized physical activities (i.e., swimming, sports teams).
While the sensory subtypes differed in regard to frequency of participation and level of involvement in home, school, and community activities, findings revealed that the effect size was greatest for involvement in community activities ($r^2_s = 0.72$). This finding is not altogether surprising, given that children with disabilities have fewer environmental supports to facilitate participation in the community than they do in home and school settings (Bedell et al., 2013). In addition to having fewer supports in this setting, perceived environmental supports and barriers have been documented to have a more significant impact on a child’s level of participation in the community than in home and school settings (Anaby et al., 2014). Another explanation for this finding may be that it is difficult for children with ASD to overcome the barrier of social demands that accompany community participation (Tint, Maughan, & Weiss, 2016). Results from other studies have shown that children in the PI and GSD subtypes demonstrate more maladaptive and socially withdrawn behaviors than children in other subtypes; this may, in part, explain the observed patterns in the present data, where children in these subtypes did not significantly differ in levels of participation (Lane et al., 2010; Tanner & Lane, unpublished manuscript).

3.5.1 Limitations and future directions

There are some limitations to the present study that should be considered. First, the child’s ASD diagnostic status was determined by parent report. We can be confident that children of participants recruited through the IAN received an independent ASD diagnosis due to the recent validation of diagnoses in the registry. However, there is less certainty about the diagnostic status for participants who responded to waiting room fliers.
and ResearchMatch emails. Thus, it is advisable that future replication of this study be conducted in a sample of children with independently confirmed ASD diagnoses.

Additionally, we hypothesize that sampling bias may have resulted in the disproportionately small number of participants belonging to the SA subtype, when the present sample is compared with other independent samples of children with ASD (Lane et al., 2014a). Sampling bias may have been introduced as a result of recruitment materials that mentioned an interest in “sensory processing” and child participation. Although the materials indicated that caregivers of children with ASD with and without sensory difficulties could complete the survey, mention of this special interest may have decreased the likelihood of response by caregivers of children in the SA subtype. Additionally, it is possible that caregivers of children in the SA subtype were not presented with the same opportunities for participation as caregivers of children in other subtypes. Due to the mild nature of their sensory features, it is possible that children in this subtype do not receive outpatient occupational, physical, or speech therapy services and as such, their caregivers did not have the opportunity to respond to waiting room fliers in local clinics. Future studies should carefully consider these aspects of recruitment in seeking to recruit more proportionate numbers of participants in the SA subtype.

Third, the present study did not control for the type of school setting (i.e., home school, private school, public school, alternative school) that participants attend. Review of the school participation section of the PEM-CY suggests that at least some items are appropriate regardless of the type of school that the child attends (i.e., completes in-class assignments, goes on field trips). However, other items may not be appropriate to
children who do not attend traditional school settings. For example, children who are home schooled are not presented with opportunities to participate in special roles at school (i.e., lunch room supervisor, student mentor), or participate in in-class group work. As such, type of school setting is an important covariate to consider for future studies, as it may have been a confounding variable in the present analysis.

Finally, there are some limitations associated with CCA that should be noted. As CCA is associational in nature, we are unable to assert any causal relationships between sensory subtypes and level of child participation. We can, however, conclude that subtype membership is associated with different frequencies of participation and levels of involvement in home, school, and community activities. Additionally, because CCA reflects the variance shared by the canonical variates (i.e., linear composites of observed variables) rather than the observed variables themselves, it is recommended that the present study be replicated with a larger sample size that would sufficiently power alternate statistical methodologies that would allow for direct comparisons between subtypes on observed variables (i.e., MANCOVA). Lastly, as a power analysis indicated that a sample size of 282 would be sufficient to detect a small effect at 80% power, there may be some concern that the present study was overpowered and subsequently resulted in increased risk for Type I error. In order to test the possible effect of a large sample size, the CCA was re-run with a random selection of 75% (n = 276) of the data. The analysis with a random subset of data yielded the same substantive interpretation as the results presented here, which lends credibility to the robustness of these findings.
3.6 Summary and Conclusion

The present study found a significant association between sensory subtypes of children with ASD and frequency of participation and level of involvement in home, school, and community activities, while controlling for household income, child age, and number of children in the household. Children in the SA subtype demonstrated the highest levels of participation, while children in the TSS subtype were reported to have the second highest levels of participation, when compared with children in the GSD subtype. Children in the PI and GSD subtypes did not significantly differ in levels of participation.

Findings of the present study may assist in improving the participation of children with ASD by alerting clinicians to aspects of participation relevant to the child’s sensory subtype to target for intervention. These findings, when combined with existing literature, have the capability to support decision making for clinicians working with children with ASD by linking each subtype with intervention targets and evidence-based interventions. Ultimately, such a clinical decision making support may lead to improved treatment outcomes for children with ASD.
Chapter 4: Identification of a subset of Short Sensory Profile items to predict sensory subtype membership in children with ASD

4.1 Abstract

*Background:* Sensory subtypes refer to groups of children with autism spectrum disorder (ASD) who demonstrate shared patterns of difficulties with sensory processing. The Short Sensory Profile (SSP) is a caregiver questionnaire that measures sensory processing and has been used to identify four sensory subtypes of children with ASD: 1) Sensory Adaptive; 2) Taste/Smell Sensitive; 3) Postural Inattentive; and 4) Generalized Sensory Difference. The Sensory Adaptive subtype is characterized by a lack of clinically meaningful sensory processing difficulties. The remaining subtypes, which demonstrate clinically meaningful levels of difficulty with sensory processing, differ from one another by their scores on the sensory domains of the SSP. A child’s sensory subtype is determined based on SSP domain z-scores, requiring caregivers to answer all 38 SSP items, which may limit the clinical utility of sensory subtyping. *Purpose:* The purpose of the present study is to improve the efficiency and utility of sensory subtype determination by identifying a subset of items from the SSP that reliably determine a child’s sensory subtype membership. *Method:* Data from a national survey of caregivers of children with ASD (n = 349), consisting of children belonging to the three subtypes with clinically meaningful sensory processing difficulties, was used in a penalized multinomial logistic regression model. *Results:* Eighteen items from the SSP were found to predict sensory
subtype membership with 94.3% raw agreement and an Adjusted Rand Index of 0.82.

Conclusions: The reduced item subset is a short and accurate method for determining sensory subtype membership for children in the Taste/Smell Sensitive, Postural Inattentive, and Generalized Sensory Difference subtypes.
4.2 Introduction

The known heterogeneity in the clinical presentation of children with autism spectrum disorder (ASD) is a barrier to the successful treatment of children with ASD, as interventions may work for some children, but not others (Bölte, 2014; Scheeren, Koot, & Begeer, 2012). As a result, there is a widespread effort to identify homogenous subtypes of children with ASD that may respond differentially from other subtypes to intervention (The Interagency Autism Coordinating Committee, 2014). Such subtypes may then inform clinical reasoning supports for clinicians working with children with ASD by linking each subtype to known areas of functional performance limitations and evidence-based interventions.

Sensory subtypes, which are groups of children with ASD who demonstrate shared patterns of responses to everyday environmental stimuli, have been proposed as one useful approach to the identification of ASD subtypes (Schaaf & Lane, 2015a). Indeed, sensory subtypes of children with ASD have been documented across multiple independent samples and have been corroborated with behavioral and neurophysiologic findings (Ausderau et al., 2014; Baranek, David, Poe, Stone, & Watson, 2006; Lane, Young, Baker, & Angley, 2010; Lane, Dennis, & Geraghty, 2011; Lane, Eldridge, Hand, Harpster, & Dennis, 2015; Schaaf & Lane, 2015b). The earliest sensory subtypes were those published by Lane and colleagues, who propose four sensory subtypes of children with ASD based on parent responses to the Short Sensory Profile (Lane et al., 2010). The Short Sensory Profile (SSP) measures behaviors associated with differences in reactivity to everyday, environmental sensory stimuli across seven sensory domains: 1) tactile; 2) taste/smell; 3) movement; 4) visual/auditory sensitivity; 5) under-responsive/seeks
sensation; 6) auditory filtering; and 7) low energy/weak (McIntosh et al., 1999). Scores on each domain are then compared to normative data from 1,200 typically developing children. Higher scores relate to more typical performance, whereas lower scores indicate that either a probable or definite difference in sensory processing is likely.

As a result of a model-based cluster analysis on the SSP, Lane et al. (2014a) posit four sensory subtypes of children with ASD: 1) Sensory Adaptive (SA); 2) Taste/Smell Sensitive (TSS); 3) Postural Inattentive (PI); and 4) Generalized Sensory Difference (GSD). The SA subtype is characterized by mild difficulties in the SSP domains of auditory filtering and under-responsive/seeks sensation; however, it is unlikely that these difficulties affect functional performance. The TSS subtype is characterized by extreme taste/smell sensitivity and the PI subtype displays extreme scores in the low energy/weak domain; both of these subtypes display clinically meaningful concerns in the auditory filtering and under-responsive/seeks sensation domains. The GSD subtype is characterized by clinically meaningful difficulties in all seven sensory domains measured by the SSP.

Based on these descriptions, performance on four SSP domains best distinguish the sensory subtypes from one another: 1) under-responsive/seeks sensation; 2) auditory filtering; 3) taste/smell sensitivity, and 4) low energy/weak. The under-responsive/seeks sensation and auditory filtering domains are hypothesized to map differences between the subtypes in severity of sensory difficulties, where those in the SA subtype have only mild difficulties, TSS and PI have intermediate difficulty, and GSD have the most difficulty. The domains of taste/smell sensitivity and low energy/weak differentiate the two intermediate severity subtypes, TSS and PI, from one another. Additionally, an
independent component analysis revealed that three of these domains were significant contributors to the underlying constructs of the SSP and the sensory subtypes (Hand et al., unpublished manuscript). Findings suggest that the underlying constructs are: 1) sensory reactivity, which is the intensity of a response to a sensory stimulus and 2) multisensory integration, which is the ability to process multiple sensory stimuli concurrently (Hand et al., unpublished manuscript). The three SSP domains that contributed most to these two constructs were taste/smell sensitivity, low energy/weak, and under-responsive/seeks sensation. Taken together, these findings suggest that it may be possible to determine sensory subtype membership with a reduced subset of items from the SSP, as some domains may be more salient to subtype determination than others.

Although the SSP is useful for identifying sensory subtypes of children with ASD, the length of this questionnaire may limit the clinical and research utility of sensory subtyping. An interdisciplinary diagnostic evaluation for a child with suspected ASD is considered best practice in clinical diagnosis and assessment (Spence, Sharifi, & Wiznitzer, 2004). The interdisciplinary team often consists of psychologists; neurologists; speech-language pathologists; audiologists; occupational and physical therapists; and special educators. These service providers work to conduct a comprehensive evaluation of the child including a formal diagnostic test (i.e., Autism Diagnostic Observation Schedule) as well as appraisal of speech and language, cognitive and adaptive behavior, sensory processing, motor coordination, family resources, behavior, and academic achievement (Filipek et al., 1999). As such, it is critical that service providers work to “maximize efficient use of time” during the evaluation process (Filipek et al., 1999, p. 456). One way that clinicians maximize efficient use of their time
is to select “short form” assessments that provide a quick and accurate estimate of the child’s functioning (Jeyakumar, Warriner, Raval, & Ahmad, 2004). Additionally, as it is strongly recommended, and sometimes necessary due to behavioral difficulties, that parents are active participants in the evaluation process, reducing the amount of time that parents spend filling out questionnaires increases the amount of time that they can be actively involved in the evaluation (Filipek et al., 1999; Tomchek & Koenig, 2016).

Similarly, in research situations, assessment brevity may be highly valuable. In online surveys, for example, it is common that caregivers complete a battery of tests. Research demonstrates that shorter duration of a web-based survey is a significant predictor of a higher response rate (Crawford, Couper, & Lamias, 2001; Deutskens, Ruyter, Wetzels, & Oosterveld, 2004; Fan & Yan, 2010). In a meta-analysis, Fan et al. (2010) found that online surveys 13 minutes or less in duration were associated with the highest response rate. Moreover, in intervention trials, it is common to collect both primary and secondary outcome measures; for example, a recent randomized controlled trial of Sensory Integration therapy for children with ASD utilized four assessments consisting of both performance-based measures and caregiver questionnaires (Pfeiffer, Koenig, Kinnealey, Sheppard, & Henderson, 2011). Given that active involvement of the parent is recommended in the assessment of a child with ASD, it may be advantageous to minimize the length of time that the caregiver spends filling out questionnaires in research evaluations (i.e., baseline assessment, outcome assessment) in addition to clinical settings (Tomchek & Koenig, 2016).

Currently, the sensory subtyping algorithm proposed by Lane and colleagues is based on SSP domain-level z-scores, requiring caregivers to answer all 38 items to
determine a child’s sensory subtype. Reducing the number of items required for subtype determination will increase the efficiency of measurement for both clinical and research settings. Thus, the objective of the present study is to determine a subset of items from the SSP that can be used to distinguish between the sensory subtypes with >90% agreement with the existing algorithm. Preliminary work utilizing item response theory methods on SSP data from children with attention-deficit hyperactivity disorder, sensory processing disorder, and ASD suggests that a subset of 4-10 items from the SSP may be sufficient for sensory subtype determination (Hand et al., 2016). However, as the sensory subtypes have not been validated in other diagnostic populations and attempts to replicate the cluster model from which the sensory subtypes were derived were less successful in heterogeneous diagnostic populations, it is necessary to explore item reduction in a sample of only children with ASD. Additionally, we hypothesize that the SSP domains of taste/smell sensitivity, low energy/weak, under-responsive/seeks sensation, and auditory filtering will be heavily represented in the reduced item subset.

4.3 Materials and Methods

4.3.1 Participants

Data for the present study were collected as part of a larger survey examining the relationship between sensory subtypes in children with ASD, child participation in daily activities, and caregiver burden. The survey used a non-experimental, cross-sectional design. Survey participants were primary caregivers, defined as the person who takes primary responsibility for the child, of children with ASD aged 5-13 years (APA, 2013). Caregivers were excluded if their child had a comorbid neurological diagnosis (i.e.,
Participants were recruited through the Interactive Autism Network (IAN), ResearchMatch, and pediatric clinics in the greater Columbus, Ohio area. The IAN and ResearchMatch are online research registries. The IAN is an autism-specific research registry, which recently authenticated the parent-report ASD diagnoses of children in their database via medical record review (Daniels et al., 2011). A total of 8,215 registrants in the IAN and 255 registrants in ResearchMatch were approached for participation in the study. Participants meeting study criteria from the IAN and ResearchMatch received an email containing a brief study description and link to the survey site. A series of three emails were sent to each potential participant to maximize survey response rate. Fliers for the present study were also posted in waiting rooms and reception areas of local pediatric occupational, physical, and speech therapy clinics. Participants received a Redbox DVD code via email as a token of appreciation for survey completion. This survey was reviewed and approved by the Institutional Review Board of The Ohio State University.

4.3.2 Measures

*Short Sensory Profile.*

The SSP (McIntosh et al., 1999) was designed to measure sensory processing in children aged 3-10 years, but has been widely used in populations of children over 10 years of age (Mangeot et al., 2001; Schoen et al., 2008; Tavassoli et al., 2016; Uljarević et al., 2016). As the SSP demonstrates convergent validity with conceptually similar measures in individuals with ASD up to 14 years of age (Tavassoli et al., 2016), and in
individuals with sensory processing difficulties, but not ASD, up to 16 years of age (Schoen et al., 2008), a broader age range of participants is reflected in the present sample. In populations of children aged 3-10 years, the internal consistency of overall and subdomain sections of the SSP reportedly ranges between $r = 0.70 - 0.90$ and discriminative validity is acceptable (> 95% in differentiating children with and without sensory impairments, McIntosh et al., 1999). The seven-domain structure of the SSP was determined via factor analysis using data collected from 1,037 typically developing children (Dunn & Brown, 1997).

### 4.3.3 Data analysis

Participants who had overall missing data rates for the SSP greater than 15% ($n = 3$), or who had 50% or more missing data from any single SSP domain ($n = 3$) were excluded from analyses due to the impact this may have on subtype determination. Additionally, participants who did not report child age were excluded from analysis, as their eligibility for the study could not be confirmed ($n = 3$).

Twenty-five participants with missing SSP values were included in the present analysis. The total number of missing items among these participants was 34, resulting in an overall missing data rate for the SSP of 0.24%. Iterative model-based imputation using robust stepwise regression was used to impute missing values for these participants with the package “VIM” in the statistical software R (Templ et al., 2015). In this type of imputation, in each step of the iteration, one variable is used as a response variable and the remaining variables serve as the regressors. The iterations continue until the estimate of the missing value stabilizes. The sensory subtype for each participant was then determined using the algorithm published by Lane and colleagues.
For additional validation, as the impact of imputing missing SSP values on sensory subtype determination has not been tested, an expert on sensory subtypes examined SSP domain z-scores for participants with missing values (n = 25) prior to imputation. By visually inspecting the z-scores, the expert identified the subtype in which they hypothesized each participant belonged. The agreement rate between the classifications based on the expert’s visual inspection and the Lane et al. (2014a) algorithm-determined subtype classification based on the imputed values was 68%. Although this is a relatively conservative level of agreement, we chose to include these participants in subsequent analyses as the missing data rate for the SSP was small (0.24%).

To identify the most salient SSP items for sensory subtype determination, a multinomial logistic regression with lasso penalty was conducted. Sensory subtype was used as the dependent variable, while item-level responses to the 38 SSP items were independent variables. Lasso regression was used for two reasons: 1) it permits the selection of independent variables (SSP items) in relation to a dependent variable, and 2) it produces a parsimonious model (Tibshirani, 1996). Lasso regression requires selection of a penalty parameter value (λ), which ranges from zero, indicating an un-penalized model, to infinity, indicating a model in which all non-intercept coefficients are shrunk to zero. To aid in the selection of a λ value, 10-fold cross-validation was conducted for 100 different, randomly-selected λ values using the statistical software R with add-on package “glmnet” (Friedman, Hastie, Simon, & Tibshirani, 2014). In 10-fold cross-validation, the data are divided into ten roughly equal parts, called folds. The model is fit omitting one of the folds and is used to predict sensory subtype membership on the
omitted fold. This process is then repeated for each of the folds. Ten-fold cross-validation results in regression weights that are optimized for predictive power in a new data set and are, therefore, likely to be generalizable beyond the sample that was used to derive the model (Arlot & Celisse, 2010). The rationale for using 10 folds, rather than a different number of folds, is that 10-fold cross-validation has been shown to be optimal for model selection (Kohavi, 1995). For each of the 100 tested λ values, a mean squared cross-validated error and regression weights were produced. For the present study, a λ value was selected to maximize model parsimony, minimize the mean squared error to maximize generalization, and maintain > 90% agreement with the classification based on the algorithm proposed by Lane and colleagues.

Intercepts and regression weights for the items that were determined relevant by the lasso regression were used to generate a new sensory subtyping algorithm. Level of agreement between sensory subtype classifications derived from this novel algorithm and those derived from the Lane et al. algorithm was assessed with raw percent agreement and the Adjusted Rand Index (ARI). The ARI is a measure of the similarity between two classification mechanisms, adjusted for chance (Hubert & Arabie, 1985). An ARI close to 1 indicates good agreement between the two classification mechanisms, while an ARI close to 0 indicates a level of agreement that would be expected by chance alone. The R packages used to determine raw percent agreement and ARI were “irr” and “fossil,” respectively (Gamer, Lemon, Gamer, Robinson, & Kendall’s, 2012; Vavrek, 2012).

4.4 Results

Due to the disproportionately small number of participants in the SA subtype (n = 20), this group was not included in the regression analysis. Therefore, the reduced
item subset generated as a result of this study differentiates the three subtypes with clinically meaningful sensory processing concerns (TSS, PI, and GSD) from one another. It does not, however, identify members of the SA subtype. Implications for this are further entertained in the discussion section. With the removal of children in the SA subtype, the total number of participants included in the multinomial logistic regression analysis was 349.

Descriptive information about the participants in the present analysis is detailed in Table 4.2. The majority of children included in the study were male (93.70%), with an average age of 10.08 years (SD = 2.52). Nearly 40% of participants were between 11 and 13 years of age. The average age of caregivers completing the surveys was 42.49 years (SD = 7.03), and 83.66% were female. The majority of caregivers were married (81.08%), and 97% had either attended or graduated from a college. Nearly 69% of caregivers were employed full or part time.
<table>
<thead>
<tr>
<th></th>
<th>All</th>
<th>TSS</th>
<th>PI</th>
<th>GSD</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>n</strong></td>
<td>349</td>
<td>137</td>
<td>78</td>
<td>134</td>
</tr>
<tr>
<td><strong>Median caregiver age</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>(IQR)</td>
<td>42.17 (37.29-46.66)</td>
<td>41.99 (38.07-47.31)</td>
<td>42.02 (37.35-45.80)</td>
<td>42.49 (36.16-46.28)</td>
</tr>
<tr>
<td><strong>Median child age</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>(IQR)</td>
<td>10.30 (7.72-11.99)</td>
<td>9.72 (7.47-12.21)</td>
<td>10.63 (8.08-12.20)</td>
<td>10.33 (8.11-12.65)</td>
</tr>
<tr>
<td><strong>% Male (child)</strong></td>
<td>93.70</td>
<td>90.51</td>
<td>96.15</td>
<td>95.52</td>
</tr>
<tr>
<td><strong>Caregiver highest level of education</strong>*</td>
<td>Some graduate school</td>
<td>Some graduate school</td>
<td>Some graduate school</td>
<td>Some college/Associates degree</td>
</tr>
<tr>
<td><strong>Median number of children in home</strong></td>
<td>2</td>
<td>2</td>
<td>2</td>
<td>2</td>
</tr>
</tbody>
</table>

*Note. *Mode. TSS = Taste/Smell Sensitive; PI = Postural Inattentive; GSD = Generalized Sensory Difference; IQR = Interquartile range
As lasso regression requires the selection of a penalty term ($\lambda$), which influences the intercepts and regression weights, we first identified an optimal $\lambda$ value using an iterative process for model selection (Figure 4.1). The process started with the selection of an arbitrary $\lambda$ value. The intercepts and regression weights corresponding to that $\lambda$ value were input into a novel algorithm used to identify sensory subtype membership based on item-level responses. Next, the level of agreement between the novel algorithm and the Lane et al. algorithm was calculated. By examining the number of items that would be included in the algorithm for a given $\lambda$ value and the level of agreement, we modified the selection of the $\lambda$ value until we reached a balance between item parsimony and agreement. Table 4.3 provides a selected range from the tested $\lambda$ values to illustrate the data used for model selection. A $\lambda$ value of 0.075 was selected by balancing model parsimony and >90% agreement with the gold standard.
Figure 4.1 Iterative process of model selection

Select penalty term

Agreement  Algorithm

Note. Penalty term = $\lambda$ value for lasso regression; Algorithm = novel algorithm developed from intercepts and regression weights for a given $\lambda$ value; Agreement = raw percent agreement and Adjusted Rand Index between the novel algorithm and the Lane et al. (2014a) algorithm.
Table 4.3 Summary of models across a range of $\lambda$ values

<table>
<thead>
<tr>
<th>$\lambda$</th>
<th>CV Mean Error (95% CI)</th>
<th># Items</th>
<th>% Agreement</th>
<th>ARI</th>
</tr>
</thead>
<tbody>
<tr>
<td>3.25e-03</td>
<td>0.31 (0.27-0.35)</td>
<td>35</td>
<td>100.00</td>
<td>1.00</td>
</tr>
<tr>
<td>4.83e-02</td>
<td>0.67 (0.63-0.72)</td>
<td>28</td>
<td>96.80</td>
<td>0.90</td>
</tr>
<tr>
<td>7.50e-02*</td>
<td>0.85 (0.81-0.90)</td>
<td>18</td>
<td>94.30</td>
<td>0.82</td>
</tr>
<tr>
<td>1.10e-01</td>
<td>0.99 (0.95-1.03)</td>
<td>14</td>
<td>89.70</td>
<td>0.70</td>
</tr>
<tr>
<td>3.41e-01</td>
<td>1.98 (1.96-2.00)</td>
<td>9</td>
<td>86.20</td>
<td>0.61</td>
</tr>
</tbody>
</table>

*Selected model. CV mean error = Cross-validated mean squared error (95% confidence interval); # Items = The number of items included in the novel algorithm; % Agreement = Raw agreement between the two algorithms; ARI = Adjusted Rand Index
Examination of regression weights corresponding to the chosen $\lambda$ value supports the face validity of this reduced-item subset (Table 4.4). In this context, a negative regression weight of an item for a subtype indicates that a higher score on that item, or a more typical behavioral response to the sensory stimulus, reduces the probability that the participant belongs to that subtype. Conversely, a positive regression weight indicates that the higher the score on a given item, the higher the probability that the participant belongs to that subtype. Results indicate that children with ASD are most likely to belong to the GSD subtype if they demonstrate low scores on a variety of items that span across multiple SSP domains. This is consistent with the description of the GSD subtype from Lane and colleagues (2014a), which indicates that children with the GSD subtype demonstrate clinically meaningful sensory processing difficulties in all SSP domains. Examination of the TSS and PI subtype regression weights reveals that scores on items from the taste/smell sensitivity and low energy/weak domains continue to differentiate these subtypes from one another. Specifically, children with higher scores on taste/smell sensitivity domain items 8-11 are more likely to belong to the PI subtype than the TSS subtype; this finding is consistent with the Lane et al. (2014a) classification schema where children in the TSS subtype demonstrate extremely low scores on taste/smell sensitivity domain items. Conversely, children with higher scores on items 31 and 32 from the low energy/weak domain are more likely to belong to the TSS subtype than the PI subtype, which parallels the original classification schema in that the PI subtype is associated with low scores in the low energy/weak SSP domain.
Table 4.4 Lasso multinomial logistic regression intercepts and regression weights showing predictors of sensory subtype membership

<table>
<thead>
<tr>
<th>Domain</th>
<th>SSP Item #</th>
<th>$\beta^{TSS}$</th>
<th>$\beta^{PI}$</th>
<th>$\beta^{GSD}$</th>
</tr>
</thead>
<tbody>
<tr>
<td>Tactile Sensitivity</td>
<td>1</td>
<td>.</td>
<td>.</td>
<td>-0.23</td>
</tr>
<tr>
<td></td>
<td>3</td>
<td>.</td>
<td>.</td>
<td>-0.02</td>
</tr>
<tr>
<td></td>
<td>5</td>
<td>.</td>
<td>.</td>
<td>-0.07</td>
</tr>
<tr>
<td></td>
<td>6</td>
<td>.</td>
<td>.</td>
<td>-0.01</td>
</tr>
<tr>
<td></td>
<td>7</td>
<td>0.10</td>
<td>.</td>
<td></td>
</tr>
<tr>
<td>Taste/Smell Sensitivity</td>
<td>8</td>
<td>.</td>
<td>0.36</td>
<td></td>
</tr>
<tr>
<td></td>
<td>9</td>
<td>.</td>
<td>0.54</td>
<td></td>
</tr>
<tr>
<td></td>
<td>10</td>
<td>.</td>
<td>0.39</td>
<td></td>
</tr>
<tr>
<td></td>
<td>11</td>
<td>.</td>
<td>0.47</td>
<td></td>
</tr>
<tr>
<td>Movement Sensitivity</td>
<td>12</td>
<td>.</td>
<td>.</td>
<td>-0.32</td>
</tr>
<tr>
<td></td>
<td>13</td>
<td>0.24</td>
<td>.</td>
<td>-0.06</td>
</tr>
<tr>
<td></td>
<td>14</td>
<td>.</td>
<td>.</td>
<td>-0.33</td>
</tr>
<tr>
<td>Under-responsive/Seeks</td>
<td>17</td>
<td>0.05</td>
<td>.</td>
<td></td>
</tr>
<tr>
<td>Auditory Filtering</td>
<td>24</td>
<td>0.02</td>
<td>.</td>
<td>-0.01</td>
</tr>
<tr>
<td>Low Energy/Weak</td>
<td>31</td>
<td>0.09</td>
<td>.</td>
<td>-0.12</td>
</tr>
<tr>
<td>Visual/Auditory Sensitivity</td>
<td>34</td>
<td>0.05</td>
<td>.</td>
<td>-0.14</td>
</tr>
<tr>
<td></td>
<td>36</td>
<td>0.03</td>
<td>.</td>
<td>-0.04</td>
</tr>
<tr>
<td><strong>Intercepts</strong></td>
<td></td>
<td><strong>-1.48</strong></td>
<td><strong>-4.53</strong></td>
<td><strong>6.01</strong></td>
</tr>
</tbody>
</table>

Note. $\beta^{TSS}$, $\beta^{PI}$, and $\beta^{GSD}$ = Regression weights for prediction of TSS, PI, and GSD subtypes, respectively.
The model yielded eighteen SSP items for the prediction of sensory subtype membership, which are described in Table 4.5. All seven sensory domains of the SSP are represented in the eighteen-item subset.
Table 4.5 Items selected by lasso multinomial logistic regression

<table>
<thead>
<tr>
<th>SSP Domain</th>
<th>Item</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Tactile Sensitivity</strong></td>
<td>1. Expresses distress during grooming […]</td>
</tr>
<tr>
<td></td>
<td>3. Avoids going barefoot, especially in sand or grass</td>
</tr>
<tr>
<td></td>
<td>5. Withdraws from splashing water</td>
</tr>
<tr>
<td></td>
<td>6. Has difficulty standing in line or close to other people</td>
</tr>
<tr>
<td></td>
<td>7. Rubs or scratches out a spot that has been touched</td>
</tr>
<tr>
<td><strong>Taste/Smell Sensitivity</strong></td>
<td>8. Avoids certain tastes or food smells […]</td>
</tr>
<tr>
<td></td>
<td>9. Will only eat certain tastes</td>
</tr>
<tr>
<td></td>
<td>10. Limits self to particular food textures/temperatures</td>
</tr>
<tr>
<td></td>
<td>11. Picky eater, especially regarding food textures</td>
</tr>
<tr>
<td><strong>Movement Sensitivity</strong></td>
<td>12. Becomes anxious or distressed when feet leave the ground</td>
</tr>
<tr>
<td></td>
<td>13. Fears falling or heights</td>
</tr>
<tr>
<td></td>
<td>14. Dislikes activities where head is upside down</td>
</tr>
<tr>
<td><strong>Under-responsive/ Seeks</strong></td>
<td>17. Becomes overly excitable during movement activity</td>
</tr>
<tr>
<td><strong>Auditory Filtering</strong></td>
<td>24. Can’t work with background noise (for example, fan, refrigerator)</td>
</tr>
<tr>
<td><strong>Low Energy/Weak</strong></td>
<td>31. Can’t lift heavy objects […]</td>
</tr>
<tr>
<td></td>
<td>32. Props to support self</td>
</tr>
<tr>
<td><strong>Visual/ Auditory Sensitivity</strong></td>
<td>34. Responds negatively to unexpected or loud noises […]</td>
</tr>
<tr>
<td></td>
<td>36. Is bothered by bright lights after others have adapted to the light</td>
</tr>
</tbody>
</table>
4.5 Discussion

To improve the efficiency of sensory subtyping, the present study sought to identify a subset of items from the SSP that can determine sensory subtype membership of children with ASD with a high degree of agreement with the existing algorithm. The findings of the present study yielded eighteen items from the SSP that can be used for distinguishing the TSS, PI, and GSD subtypes from one another. In concordance with our hypothesis, we found that all items from the taste/smell sensitivity domain were included in the item subset, and that higher scores on these items predicted membership to the PI subtype. Some items from the low energy/weak, under-responsive/seeks sensation, and auditory filtering domains were included in the item subset, however there were fewer items included from these domains than anticipated. This finding is likely due to the use of lasso regression, which unceremoniously selects one variable from a group of highly correlated predictors (Tibshirani, 1996). Further, the 18-item subset effectively represents all seven SSP sensory domains, which suggests that while the domains of under-responsive/seeks sensation, auditory filtering, taste/smell sensitivity, and low energy/weak best highlight differences between the subtypes, all seven SSP domains contribute unique information that is necessary for successful subtype identification. One possible explanation for this is that the items included from other domains may also serve as severity classifiers of overall sensory processing, which help to differentiate between the subtypes. For example, four of the items from the tactile sensitivity domain (distress during grooming; avoids going barefoot; difficulty standing in line; and withdraws from splashing water) and two items from the movement sensitivity domain (distress when feet leave the ground; and dislikes activities where head is upside down) were significant
predictors of GSD subtype membership. Substantively, this indicates that children with high scores on these items, which indicate a more typical behavioral response, are not likely to demonstrate the severity of sensory processing difficulties that is characteristic of the GSD subtype.

While we were able to substantially reduce the number of items needed for sensory subtype determination, we were unable to reliably predict sensory subtype membership with 10 or fewer SSP items. As with all short forms, there is a trade-off between questionnaire length and the breadth and precision of measurement (Ware & Sherbourne, 1992). The loss of predictive accuracy for models with less than 18 items was judged to be too great, as we were unable to achieve > 90% agreement with the existing algorithm. The 18-item subset, however, achieved a percent agreement of 94.3% with the existing algorithm, which is comparable to other widely used short forms, such as the Short Form 12 Health Survey (Ware, Kosinski, & Keller, 1996). In choosing between the SSP and the 18-item subset, it is important to consider that the SSP provides a more global understanding of a child’s sensory processing, whereas the 18-item subset was designed for the sole purpose of differentiating the three subtypes with clinically meaningful sensory concerns from one another. Consequently, the choice of using this reduced item subset is more justified in research or clinical situations where: 1) membership to the SA subtype has been ruled out; 2) there are constraints on questionnaire length or administration time; and 3) the researcher or clinician seeks to identify a child’s sensory subtype, rather than to obtain a broader view of overall sensory processing.
4.5.1 Exclusion of the SA subtype

In the present study, we were unable to reliably detect membership to the SA subtype. We hypothesize that this is due to the low number of participants (n = 20) belonging to that subtype. In analyses conducted prior to the decision to exclude those participants, regardless of the selected λ value, the algorithm failed to identify any participants as belonging to the SA subtype, incorrectly classifying them as belonging to the TSS or PI subtypes. It is possible that with a more proportionate number of participants in the SA subtype, the proposed algorithm could be refined to allow for accurate classification of members of all four sensory subtypes. We would anticipate that the SA subtype would be characterized by very large, positive regression weights across a wide range of items.

4.5.2 Implications for clinical and research utility

The novel algorithm proposed here reduces the number of SSP items that parents must answer for subtype determination by over 50%. As the SSP is estimated to take 10 minutes to complete (McIntosh et al., 1999), this reduced version may take less than five minutes for parent completion, which serves to improve the clinical and research utility of sensory subtyping. Clinically, this reduced item subset may be particularly useful in situations where time is limited, such as an interdisciplinary diagnostic evaluation of a child with suspected ASD (Filipek et al., 1999). Figure 4.2 provides a graphic illustration of the breadth and depth of assessment by such an interdisciplinary team. Due to the extent of testing to be done, reducing the duration of time that the caregiver spends completing the SSP by five minutes makes it possible for the parent to re-allocate that time to providing developmental histories or discussing concerns with service providers.
Moreover, it is strongly recommended that parents be intimately involved in the evaluation process for children with ASD (Tomchek & Koenig, 2016). The subset of SSP items identified in this study, which reliably and efficiently determine a child’s sensory subtype, may both: 1) aid clinicians in the pursuit of maximizing efficiency, and 2) provide parents with additional time that they are able to spend actively engaging in the evaluation via discussions, observations, and facilitating their child’s participation.
Figure 4.2 Interdisciplinary evaluation process for a child with suspected ASD

Note. Schematic derived from practice recommendations from Filipek et al., 1999; SIPT = Sensory Integration and Praxis Test
In addition to improving clinical usefulness, item reduction also has value for future research on sensory subtypes in children with ASD. There is an established base of literature that indicates that shorter survey length is associated with increased response rate, quality of responses, and survey completeness (Deutskens et al., 2004; Fan & Yan, 2010; Haunberger, 2011). Additionally, the proportion of survey questions that the respondent perceives as burdensome (i.e., time consuming, cognitively taxing) reduces likelihood of survey completion and follow-up survey completion (Haunberger, 2011). This may have specific implications when researchers desire to recruit a diverse sample, including individuals with lower health literacy, as individuals with lower health literacy may perceive a greater proportion of questions to be burdensome. As such, researchers seeking to conduct large surveys may wish to consider use of the reduced item subset as one method to aid in the minimization of survey length. Appendix A provides an example that illustrates how clinicians or researchers would determine a child’s sensory subtype by using this 18-item subset.

4.5.3 Limitations and future directions

Results of the present study should be considered in light of some limitations. First, the 18-item subset does not identify members of the SA subtype due to the disproportionately small number of participants belonging to the SA subtype in comparison to other independent samples of children with ASD. In the present sample, 20 participants out of 369 (5.42%) were identified as belonging to the SA subtype, while in other independent samples, the SA subtype has been documented to represent 36.84% of children with ASD (Lane et al., 2014a). We hypothesize that the reason for this may be due to sampling bias introduced by mention of an interest in “sensory processing” in
recruitment materials. Although the fliers and emails indicated that caregivers of children with ASD with and without sensory difficulties could participate, mention of this special interest may have decreased the likelihood of response by caregivers of children in the SA subtype. Additionally, it is possible that the use of clinical recruitment sites resulted in caregivers of children in the SA subtype lacking the same opportunities for participation as caregivers of children in other subtypes. Due to the mild nature of their sensory difficulties, it is possible that children in this subtype do not receive outpatient occupational, physical, or speech therapy services. As such, their caregivers may not have had the opportunity to respond to waiting room fliers in local clinics. Future studies on sensory subtypes in children with ASD should carefully consider these aspects of recruitment in seeking to recruit more proportionate numbers of children in the SA subtype. Because this 18-item subset does not identify members of the SA subtype, it is important that clinicians and researchers who wish to use this reduced item subset first rule out SA subtype membership. As children who are categorized in the SA subtype do not demonstrate functional performance limitations as a result of underlying difficulties with sensory processing, direct observation of the child and/or interviews with caregivers may be sufficient for ruling out this subtype classification.

Second, the ASD diagnostic status of children in the sample was determined by parent report. While we can be confident that children of participants recruited through the IAN received an independent ASD diagnosis due to the recent validation of diagnoses in the registry, there is less certainty with regard to participants responding to waiting room fliers and ResearchMatch emails. Therefore, it is advisable that future replication of
this study be conducted in a sample of children with independently confirmed ASD diagnoses.

Lastly, there could be some concern that the model generated as a result of this analysis is specific to the participants in our study. Careful consideration was taken in the analytical approach to maximize generalizability, such as the use of 10-fold cross-validation, which generates regression weights that are optimized for prediction in novel populations. However, it is possible that the participants in the present sample may differ in some way from other samples or the population of all children with ASD. Thus, testing the classification schema based on the 18-item subset in an independent population is advisable.

4.6 Summary and Conclusion

Sensory subtypes have important implications for the development of decision-making supports for clinicians and researchers working with children with ASD by linking subtypes with performance limitations and evidence-based interventions that may be effective for that subtype. The present study sought to improve the efficiency of sensory subtyping by reducing the number of items that caregivers must answer to determine a child’s subtype. Results yielded a subset of 18 items from the SSP that can identify children belonging to the TSS, PI, and GSD subtypes with 94.6% agreement with the algorithm based on domain z-scores for all 38 SSP items. This reduced item subset may prove useful in clinical and research situations where the goal is to determine subtype membership and there is concern about administration time or questionnaire length.
Chapter 5: Implications of Sensory Subtyping in ASD on Occupational Therapy

Treatment and Research

5.1 Summary of Findings

This body of work represents the results of three research projects focused on expanding our understanding of differences between sensory subtypes of children with autism spectrum disorder (ASD) in caregiver burden and child participation, as well as improving the efficiency of the sensory subtyping algorithm. Results of the analysis of the relationship between level of caregiver burden and sensory subtype membership of the child revealed that caregiver mental and physical health related quality of life (HRQOL) did not, but caregiver strain did, vary as a function of the child’s sensory subtype. Additionally, child participation in home, school, and community activities was found to vary between sensory subtypes. Specifically, we found that, when compared to the Generalized Sensory Difference (GSD) subtype, children in the Sensory Adaptive (SA) subtype were associated with the highest levels of participation and caregivers of these children reported the lowest levels of strain. Child membership in the Taste/Smell Sensitive (TSS) subtype was associated with an intermediate level of caregiver strain and limitations in child participation. Caregivers of children in the Postural Inattentive (PI) and GSD subtypes did not significantly differ in level of caregiver strain or child participation. These findings may, in part, be explained by the proposed theoretical
underpinnings of the sensory subtypes whereby children in both the PI and GSD subtypes demonstrate difficulties with multisensory integration.

Results from the analysis aimed at reducing the number of items necessary for sensory subtype determination indicated that 18 items from the Short Sensory Profile (SSP) can predict sensory subtype membership for the TSS, PI, and GSD subtypes with 94.3% agreement with the existing algorithm (Lane et al., 2014a). This novel sensory subtyping algorithm based on the reduced item subset, however, is not able to determine membership to the SA subtype. The authors attribute this to the disproportionally low sample size for this subtype, which may be due to sampling bias.

5.2 Limitations

Limitations for this body of work include that the survey methodology relied on caregiver report for ASD diagnosis and that sampling bias may have resulted in a low response rate from caregivers of children in the SA subtype. Additionally, we did not collect information about the type of school that the children with ASD attend, which may have had implications for the findings of the study examining participation as a function of sensory subtype (Chapter 3). The analyses used to explore relationships between caregiver burden, child participation, and sensory subtype membership (Chapter 2 and Chapter 3) were associational in nature; as such, causal inferences could not be drawn. Finally, the sample characteristics for this body of work indicate that respondents were, on average, highly educated, married women, who work full or part time and come from a high socioeconomic status. It is possible, therefore, that this sample differs from the total population of caregivers of children with ASD, and while the 18-item subset for sensory subtype determination (Chapter 4) was built for maximal predictive power, these
items may not generalize to other populations of children with ASD (i.e., children from lower socioeconomic statuses, or who have parents with less formal education).

5.3 Clinical Implications

5.3.1 Interventions for caregiver burden

While we did not find that caregiver self-reported HRQOL varied between caregivers of children in different subtypes, existing literature demonstrates that caregivers of children with ASD have poorer mental and physical HRQOL than caregivers of typically developing children and children with other developmental disabilities (Khanna et al., 2011; Allik, Larsson, & Smedje, 2006; Eapen & Guan, 2016). These findings suggest that caregivers of children with ASD in all four sensory subtypes would benefit from routine screening for level of caregiver burden and proactive measures to improve HRQOL such as improving social support and reducing maladaptive coping strategies. Higher perceived social support has been associated with fewer physical complaints and more adaptive cortisol patterns, suggesting that social support is an advantageous coping mechanism for caregivers of children with ASD (Lovell, Moss, & Wetherell, 2012). Additionally, reducing maladaptive coping behaviors and increasing positive coping strategies, such as problem solving and accepting responsibility, may have mediating effects on the relationship between stress and QOL in caregivers of children with ASD (Eapen & Guan, 2016).

Findings that caregivers of children in different subtypes varied in level of reported strain may alert clinicians to aspects of caregiver strain relevant to the child’s sensory subtype to target for intervention. Specifically, caregivers of children in the PI and GSD subtypes may be in the most need of interventions and services targeted at
alleviating caregiver strain. One intervention that may be effective at reducing caregiver strain is the utilization of respite care services. Indeed, utilization of respite care services have been associated with significant reductions in stress of caregivers of children with ASD (Cowen & Reed, 2002; Harper, Dyches, Harper, Roper, & South, 2013). In addition to reducing caregiver stress, respite care was reported to be the most effective service for reducing family and financial stress, suggesting that this intervention is highly effective for caregivers and the family unit as a whole (Ruble & McGrew, 2007).

Additionally, clinicians may wish to consider involving caregivers of children in the PI and GSD subtypes who experience high levels of strain in the treatment of the child with ASD (Karst & Hecke, 2012). Clinicians can then teach the caregiver strategies to expand their interactions with their child to foster strengthened parent-child relationships; this has been demonstrated to lead to improved outcomes for both caregivers and their children including reduced caregiver stress, increased insight into their child, and greater child generalization of skills (Brookman-Frazee & Koegel, 2004; Matson, Mahan, & Matson, 2009; McConachie & Diggle, 2007). Empowering caregivers with these skills and reinforcing parent-child relationships may reduce the frequency and intensity of negative events (i.e., missing work, interruption of personal time, and disruption of family routines due to child emotional or behavior problems) and lessen the financial burdens that contribute to perceived objective strain (Matson, Mahan, & Matson, 2009). Additionally, this approach may alleviate some negative feelings that the caregiver has toward the child with ASD that contribute to subjective externalized strain.
5.3.2 Interventions for participation

Similarly, our finding that children in different subtypes varied in their frequency of participation and level of involvement in home, school, and community activities may function to alert clinicians to aspects of participation relevant to the child’s sensory subtype to target for intervention. Specifically, children in the PI and GSD subtypes may be in the most need of interventions and services targeted at improving participation in home, school, and community activities. Specific activities that are captured in the home participation section of the Participation and Environment Measure- Children and Youth (PEM-CY), which may serve as targets for intervention to improve home participation, include indoor play and games, personal care, and household chores (Coster et al., 2014). Evidence suggests that the use of Social Stories™ may effectively improve game play skills in children with ASD (Tanner et al., 2015). Interventions that have been shown to improve independence in activities of daily living, such as personal care, for children with ASD include the Cognitive Orientation to Occupational Performance (CO-OP) model and a manualized version of Ayres’ Sensory Integration (Weaver, 2015). Additionally, the use of a contextual parent-coaching intervention, self-prompting personal digital assistants, and the CO-OP model have been shown to increase independence in household management tasks (Weaver, 2015). As such, clinicians working with children belonging to the PI and GSD subtypes may wish to consider incorporating these interventions into their treatment plans.

The school participation section of the PEM-CY measures the frequency and level of involvement a child has in activities such as classroom tasks, school-sponsored teams or clubs, and socializing with peers outside of class (Coster et al., 2014). Recess
interventions consisting of environmental and social supports such as peer prompting, teacher-led recess activities, and vibrating pagers as a tactile cue, are promising approaches for improving school participation for children with ASD (Lang et al., 2011). These approaches have been associated with improved social initiation, turn taking, and group play. Other techniques to improve school participation and performance include applied yoga, exercise, and visual supports, although the evidence to support these interventions is emerging (Weaver, 2015).

Community participation involves activities such as neighborhood outings, organized physical activities, social interaction in the community, and working for pay (Coster et al., 2014). To improve the community participation of children with ASD, clinicians may wish to consider group-based community activities, such as water exercise swimming programs, which have been shown to improve social interactions of high-functioning children with ASD (Pan, 2010). Additionally, leisure groups have been shown to decrease the need for leisure support, improve leisure engagement patterns, and increase leisure satisfaction in high functioning young adults with ASD (Palmen & Korzilius, 2011). In regard to paid employment, there is a moderate level of evidence to support the use of interventions including video-modeling, visual prompts, and self-management strategies to improve independence of individuals with ASD in work tasks (Weaver, 2015). Overall, evidence for interventions to improve community participation is limited in comparison to home and school environments. As the results of our study suggest that this is the area in most need of intervention for children in the PI and GSD subtypes, and to a lesser extent the TSS subtype, future research aimed at developing interventions to improve community participation is warranted.
Finally, it is known that many factors are associated with a child’s participation including social attitudes, communication, interpersonal relationships, and behavioral challenges (Askari et al., 2014). Thus, interventions aimed at improving these areas may result in improved participation. A recent systematic review found strong evidence that social skills groups, the Picture Exchange Communication System, joint attention interventions, and parent-mediated strategies can improve communication and social participation of children with ASD (Tanner et al., 2015). Further, interventions that are known to reduce challenging behaviors, such as the use of behavioral techniques and self-management, may also result in improved participation of children with ASD.

### 5.3.3 Development of a clinical decision-making framework

The findings of the present series of studies, along with existing literature, allow for the creation of a clinical decision-making framework for occupational therapy clinicians working with children with ASD. Currently, clinical reasoning support for clinicians working with children with ASD comes from Occupational Therapy (OT) Practice Guidelines for Individuals with ASD, published by the American Occupational Therapy Association (Tomchek & Koenig, 2016). The OT practice guidelines for ASD are based on four recent systematic reviews that examined the effectiveness of occupational therapy interventions for individuals with ASD (Kuhaneck et al., 2015; Tanner et al., 2015; Watling & Hauer, 2015; Weaver, 2015). The systematic reviews consisted of evidence Levels I, II, or III, which are systematic reviews or meta-analyses of randomized controlled trials (RCTs), at least one well-designed RCT, or well-designed quasi-experimental designs, respectively (Gliner, Morgan, & Leech, 2009 pg. 384). In the event that no Level I-III evidence could be identified, Level IV evidence, well-
designed case-control or cohort studies, were considered. The authors of each review judged the strength of the evidence for each intervention as strong, moderate, or weak (U.S. Preventive Service Task Force, 2013). Evidence for an intervention was considered to be strong if there were consistent positive findings from well-conducted studies with at least two RCTs. Evidence for an intervention was moderate if there was one RCT with positive outcomes, or two or more studies with lower levels of evidence with positive outcomes, or inconsistent findings across individual studies. Lastly, evidence was considered limited for interventions with no RCTs with positive outcomes, or studies with lower levels of evidence with inconsistent findings. While the OT practice guidelines rate the strength of the evidence for the interventions included in the systematic reviews, they do not, however, indicate for which individuals with ASD the interventions may be the most effective. The clinical decision-making framework proposed here builds upon the OT practice guidelines by linking characteristics of the child (i.e., sensory subtypes) to areas of functional performance limitations, and then to evidence based interventions.

The framework begins with a comprehensive evaluation (adapted from Alison Lane, personal communication; Figure 5.1). Parent report should be gathered via interviews or questionnaires, which may include the SSP. The clinician should also have the child perform functional activities that allow for clinical observation of the child’s developmental level as well as the impact that sensory features may have on engagement in activities. Administration of standardized assessments is also recommended, as it may provide insight into the child’s functional level when compared with a normative population. Lastly, if possible, the clinician should observe the child in a natural setting,
such as the home or classroom. While observing the child in the natural setting, the clinician should assess the influence of sensory aspects of the environment (i.e., noise, lighting, visual stimuli, smells etc.) on the child’s ability to participate in meaningful activities (i.e., social participation, attention to task, following directions, etc.).

Results of the assessment will allow the clinician to determine whether or not the child’s sensory differences interfere with functional performance on a daily basis. If the clinician finds that sensory features do not underlie the child’s difficulties (i.e., reason for occupational therapy referral), the clinician should consider alternate treatment paradigms that may be more appropriate for addressing the child’s needs. If sensory differences do contribute to daily performance limitations, the clinician should proceed to determining the child’s sensory subtype. If the SSP has not already been administered, the clinician may choose at this point to administer either the 38-item SSP or the 18-item subset to determine in which of the three subtypes with clinically meaningful sensory processing concerns the child best fits.
Figure 5.1 Clinical decision-making framework

Note. TSS = Taste/Smell Sensitive; PI = Postural Inattentive; GSD = Generalized Sensory Difference
**Sensory subtype profiles**

Comprehensive profiles for each of the four sensory subtypes of children with ASD were developed by combining findings of this body of work with existing literature and are described below. Once the child’s sensory subtype is determined, clinicians can consult the pathway for that subtype. The subtype-specific pathways guide the clinician from known functional performance limitations of that subtype to evidence-based interventions. Evidence-based interventions were primarily pulled from the four recent systematic reviews that contributed to the OT practice guidelines, as well as the OT practice guidelines themselves (Kuhaneck et al., 2015; Tanner et al., 2015; Watling & Hauer, 2015; Weaver, 2015; Tomchek & Koenig, 2016). While many occupational therapy-relevant interventions were covered in these reviews, only those that demonstrated moderate to strong evidence of efficacy were selected for inclusion in the clinical decision-making framework, unless otherwise noted. Readers are instructed to review the systematic review articles or the OT Practice Guidelines for Individuals with ASD for detailed descriptions of each of the evidence-based interventions.

**Sensory Adaptive**

The SA subtype is characterized by mild difficulties in auditory filtering as well as some under-responsive and seeking behaviors (Lane et al., 2014a). These difficulties, however, are not of sufficient magnitude to influence functional performance. In other words, any functional difficulties that children in the SA subtype may present with are not likely to be underpinned by a sensory processing difficulty. As a result, if a thorough evaluation indicates that a child best fits in the SA subtype, the clinician may wish to determine the child’s sensory strengths and identify environmental or task modifications
that can be used to support the child, and/or another intervention paradigm should be chosen. Examples of possible environmental supports that capitalize on sensory strengths may be the use of a visual schedule for a child who is strong in visual processing, or use of a therapy ball in place of a traditional chair in the classroom for a child whose attention improves with vestibular input. It is important to acknowledge, however, that research evidence for such sensory-based environmental modifications is inconclusive (Tomchek & Koenig, 2016). As a result, it is critical for the clinician to carefully monitor the child’s progress and discontinue if the intervention is deemed ineffective.

*Taste/Smell Sensitive*

The TSS subtype is characterized by challenges with sensory reactivity that manifest as extreme taste/smell sensitivity and clinically meaningful difficulties in auditory filtering, under-responsiveness, and sensory seeking behaviors (Lane et al., 2014a). Children in the TSS subtype tend to have severe communication impairments and are picky eaters (Lane et al., 2010, 2014b). They often display increased maladaptive behaviors and perform poorly in daily living skills (Lane et al., 2010; Tanner & Lane, unpublished manuscript). Additionally, children in the TSS subtype are more likely to have somatic complaints such as fatigue, aches, nausea, headaches, stomach or eye problems (Tanner & Lane, unpublished manuscript). Findings from the present body of work indicate that children in the TSS subtype demonstrate significantly higher levels of participation in home, school, and community activities than children in the GSD subtype. Additionally, caregivers of children in the TSS subtype demonstrate significantly less objective, subjective externalized, and subjective internalized strain than caregivers of children in the GSD subtype. Preliminary examination of
neurophysiologic function revealed that children in the TSS subtype demonstrate heightened response to the onset of a novel auditory stimulus and did not habituate to that stimulus over time (Lane et al., 2015). Children in this subtype have also been documented to have lower heart rate variability at baseline and during sensory stimulation, which suggests attenuated Parasympathetic Nervous System (PNS) activation resulting in an increased fight or flight response (Schaaf & Lane, 2015b). Characteristics that are indicative of children in the TSS subtype were used to create a list of possible treatment priorities that were linked to evidence-based interventions (Figure 5.2).
Figure 5.2 Taste/Smell Sensitive framework

**Step 1: Determine treatment priorities.**
Answer questions A-D. For those questions answered “yes,” proceed to Step 2.

- A) Does picky eating significantly limit the child's participation in family mealtime and/or nutritional intake?
- B) Does the child have significant delays in daily living skills for age?
- C) Does the child have significant communication impairments?
- D) Does the child have maladaptive behaviors that interfere with function?

**Step 2: Select EBP(s).**
For those questions answered “yes” in Step 1, review EBP list and select an intervention. Refer to Appendix 1 for summaries of EBPs.

**Step 3: Intervention.**
Deliver intervention and monitor intervention effectiveness. Change to another EBP if intervention is deemed ineffective for child.

- First, rule-out medical issues that could account for picky eating
  - Sensory-based interventions (i.e., Beckman sensory motor protocol)*
  - Behaviorally-based interventions*1
  - CO-OP Model*1
- Intensive clinic-based ASI1
- CO-OP Model1
- Cognitive behavioral approaches2
- Parent coaching*1
- PECS3
- Naturalistic behavioral interventions3
- Parent-mediated interventions
- Imitation training1
- Joint attention training3
- Developmental interventions*3
- Behavioral techniques3
- Daily school-based yoga*4
- Multisensory interventions*4

**Note.** *Evidence is limited. EBP = Evidence-based practice; 1) Weaver, 2015; 2) Tomchek & Koenig, 2016; 3) Tanner et al., 2015; 4) Watling & Hauer, 2015
**Postural Inattentive**

The PI subtype is characterized by difficulties with multisensory integration which manifest as deficits in postural control, endurance, and auditory filtering, as well as under-responsiveness, and sensory seeking behaviors (Lane et al., 2014a). Children in this subtype may demonstrate increased maladaptive behaviors and reduced attention to task (Lane et al., 2010; Tanner & Lane, unpublished manuscript). Additionally, children in the PI subtype may display more withdrawn behaviors (i.e., shyness, sulking, staring, sadness) as well as somatic complaints, and emotional reactivity (Tanner & Lane, unpublished manuscript). Moreover, findings presented in this body of work suggest that children in this subtype do not significantly differ from children in the GSD subtype in participation in home, school, and community activities. Additionally, caregivers of children in the PI subtype do not significantly differ from caregivers of children in the GSD subtype in levels of objective, subjective externalized, and subjective internalized strain. Based on the multisensory integration difficulties that are characteristic of children in the PI subtype, we hypothesize that these children may be especially good candidates for Ayres Sensory Integration® (ASI) due to the unique focus of ASI on the neurophysiological connections between sensation, motion, and behavior (Tomchek & Koenig, 2016). Future work to test this hypothesis with an intervention trial is warranted.

Characteristics that are associated with children in the PI subtype were used to create a list of possible treatment priorities that were linked to evidence-based interventions (Figure 5.3).
**Figure 5.3 Postural Inattentive framework**

**Step 1: Determine treatment priorities.**
Answer questions A-G. For those questions answered “yes,” proceed to Step 2.

- **A)** Does the child have difficulties with attention that interfere with engagement in daily activities?  
  - If Yes

- **B)** Does the child have maladaptive behaviors that interfere with function?  
  - If Yes

- **C)** Does the child demonstrate withdrawn behaviors (i.e., shyness, sulking) that limit social participation?  
  - If Yes

- **D)** Does the child have significant limitations in home participation?  
  - If Yes

- **E)** Does the child have significant limitations in school participation?  
  - If Yes

- **F)** Does the child have significant limitations in community participation?  
  - If Yes

- **G)** Does the child's caregiver report experiencing elevated levels of strain?  
  - If Yes

**Step 2: Select EBP(s).**
For those questions answered “yes” in Step 1, review EBP list and select an intervention. Refer to Appendix 1 for summaries of EBPs.

- **Multisensory interventions**
- **Interventions aimed at teaching the child how to self-regulate arousal level with sensory strategies (Alert Program, Zones of Regulation)**
- **Physical activity for academic engagement**

- **Behavioral techniques**
- **Multisensory interventions**
- **Daily school-based yoga**

- **Group-based interventions (UCLA PEERS, Skillstream)**
- **Collaborative tasks (LEGO, exercise)**
- **Computer-based interventions**

- **Social stories for home leisure participation**
- **CO-OP Model for home leisure participation**
- **Contextual parent-coaching for participation in ADLs and IADLs**

- **Recess interventions for social interaction and play**
- **Yoga to improve classroom behavior**
- **Exercise to improve classroom behavior**
- **Visual supports to improve classroom behavior**

- **Group-based community activities**
- **Leisure groups**
- **Video-modeling for supported employment**
- **Visual prompts for supported employment**
- **Self management strategies for supported employment**

- **Improve social support**
- **Parent involvement in treatment**
- **Connect with respite care services**
- **Parent coaching to improve confidence and self-efficacy**
- **Relaxation, mindfulness, and stress-focused interventions**

**Step 3: Intervention.**
Deliver intervention and monitor intervention effectiveness. Change to another EBP if intervention is deemed ineffective for child.

Generalized Sensory Difference

Children classified in the GSD subtype demonstrate difficulties in both sensory reactivity and multisensory integration. Specifically, this subtype is characterized by clinically meaningful concerns with sensitivity to: tactile, taste/smell, visual, auditory, and movement sensory stimuli (Lane et al., 2014a). Additionally, children in this subtype display auditory filtering deficits, low energy and weakness, under-responsiveness and sensory-seeking behaviors. Children in the GSD subtype may demonstrate more maladaptive behaviors, emotional reactivity, and somatic complaints (Tanner & Lane, unpublished manuscript). Moreover, withdrawn behaviors and poor social coping may also be observed in children in the GSD subtype. Findings of this dissertation revealed that caregivers of children in the GSD subtype are associated with significantly higher levels of objective, subjective externalized, and subjective internalized strain than caregivers of children in the SA and TSS subtypes. Additionally, children in the GSD subtype, when compared with children in the SA and TSS subtypes, demonstrate significantly greater limitations in participation in home, school, and community activities. Preliminary neurophysiologic findings suggest that children in this subtype display reduced PNS activation as well as a heightened response to the onset of a novel auditory stimulus (Lane et al., 2015; Schaaf & Lane, 2015b). Similarly to the PI subtype, children in the GSD subtype are hypothesized to be good candidates for ASI; this hypothesis has not yet been tested, however. Characteristics that are associated with children in the GSD subtype were used to identify possible treatment priorities for clinicians to consider when working with children in the GSD subtype that were linked to evidence-based interventions (Figure 5.4).
Figure 5.4 Generalized Sensory Difference framework

**Step 1: Determine treatment priorities.**
Answer questions A-G. For those questions answered “yes,” proceed to Step 2.

- **A) Does the child have maladaptive behaviors that interfere with function?**
  - If Yes
  - Behavioral techniques
  - Multisensory interventions

- **B) Does the child demonstrate withdrawn behaviors (i.e., shyness, sulking) that limit social participation?**
  - If Yes
  - Group-based interventions (UCLA PEERS, Skillstream)
  - Collaborative tasks (LEGO, exercise)
  - Computer-based interventions

- **C) Does the child demonstrate high levels of emotional reactivity that interfere with function?**
  - If Yes
  - ASI
  - Multisensory interventions
  - Environmental modifications

- **D) Does the child have significant limitations in home participation?**
  - If Yes
  - Social stories for home leisure participation
  - COOP Model for home leisure participation
  - Contextual parent-coaching for participation in ADLs and IADLs

- **E) Does the child have significant limitations in school participation?**
  - If Yes
  - Recess interventions for social interaction and play
  - Yoga to improve classroom behavior
  - Exercise to improve classroom behavior
  - Visual supports to improve classroom behavior

- **F) Does the child have significant limitations in community participation?**
  - If Yes
  - Group-based community activities
  - Leisure groups
  - Video-modeling for supported employment
  - Visual prompts for supported employment
  - Self-management strategies for supported employment

- **G) Does the child’s caregiver report experiencing elevated levels of strain?**
  - If Yes
  - Improve social support
  - Parent involvement in treatment
  - Connect with respite care services
  - Parent coaching to improve confidence and self-efficacy
  - Relaxation, mindfulness, and strength-focused interventions

**Step 2: Select EBP(s).**
For those questions answered “yes” in Step 1, review EBP list and select an intervention. Refer to Appendix 1 for summaries of EBPs.

**Step 3: Intervention.**
Deliver intervention and monitor intervention effectiveness. Change to another EBP if intervention is deemed ineffective for child.

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5.4 Research Implications

Future research should extend the work of this document through replication of this survey in a population with independently confirmed ASD diagnoses and with larger numbers of participants in the SA subtype. With a more proportionate number of participants belonging to the SA subtype, it may be possible to refine the proposed 18-item subset so that it will reliably detect members of the SA subtype in addition to the TSS, PI, and GSD subtypes. Additionally, studies seeking to elucidate the mechanisms by which sensory subtype membership is related to caregiver strain and child participation are advisable. For example, qualitative studies could evaluate differences in themes reported by caregivers of children in different subtypes. Data from this dissertation could also be used to explore potential relationships between caregiver strain and child participation in future analyses, as this may yield additional insight into the relationship between sensory subtypes, caregiver strain, and child participation. Lastly, it will be important moving forward to test for differential intervention effectiveness in children belonging to different sensory subtypes via intervention trials. Doing so may allow for further refinement of the clinical decision making framework by suggesting specific interventions for each subtype that are not only evidence based, but that are demonstrated to be maximally effective for children belonging to a given subtype.
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Appendix A: Illustration of Novel Sensory Subtyping Algorithm

To illustrate the use of the novel algorithm, we will consider a string of answers to the 18 SSP items. The participant’s responses are denoted in the column X. Regression weights from the logistic regressions predicting TSS, PI, and GSD subtype membership are found under the columns $\beta^{\text{TSS}}$, $\beta^{\text{PI}}$, and $\beta^{\text{GSD}}$, respectively and are rounded to two decimal places for ease of illustration. Items marked with a “.” are not significant predictors for that subtype.
**Step 1.** The participant’s responses (X) are multiplied by the regression weight (β) for the corresponding item. The products of the item response and the regression weight (X*β) are indicated in the yellow columns of the table.

<table>
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<th>Item</th>
<th>X</th>
<th>βTSS</th>
<th>X*βTSS</th>
<th>βPI</th>
<th>X*βPI</th>
<th>βGSD</th>
<th>X*βGSD</th>
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**Step 2.** Next, the products for each subtype are summed.

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<th>$X\cdot\beta^{TSS}$</th>
<th>$\beta^{PI}$</th>
<th>$X\cdot\beta^{PI}$</th>
<th>$\beta^{GSD}$</th>
<th>$X\cdot\beta^{GSD}$</th>
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</table>

Subtype Sum | 1.92 | 4.50 | -3.308
**Step 3.** The subtype sum (from Step 2) is added to the respective intercept to yield a total sum for each subtype. Note that the intercepts for the TSS and PI subtypes are negative.

<table>
<thead>
<tr>
<th>Subtype</th>
<th>TSS</th>
<th>PI</th>
<th>GSD</th>
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</thead>
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<td>Subtype Sum</td>
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<td>4.50</td>
<td>-3.308</td>
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<tr>
<td>Intercept</td>
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</table>
**Step 4.** The sums obtained at the end of Step 3 are used to determine probabilities of membership to each sensory subtype. The probabilities that represent the likelihood that the child belongs to each sensory subtype are calculated with the following formulae where $\text{Prob}^{\text{TSS}}$, indicates the probability of membership to the TSS subtype, and so on.

\[
\text{Prob}^{\text{TSS}} = \frac{e^{\Sigma_{\text{TSS}}}}{e^{\Sigma_{\text{TSS}}} + e^{\Sigma_{\text{PI}}} + e^{\Sigma_{\text{GSD}}}}
\]

\[
\text{Prob}^{\text{PI}} = \frac{e^{\Sigma_{\text{PI}}}}{e^{\Sigma_{\text{TSS}}} + e^{\Sigma_{\text{PI}}} + e^{\Sigma_{\text{GSD}}}}
\]

\[
\text{Prob}^{\text{GSD}} = \frac{e^{\Sigma_{\text{GSD}}}}{e^{\Sigma_{\text{TSS}}} + e^{\Sigma_{\text{PI}}} + e^{\Sigma_{\text{GSD}}}}
\]

For the current example, we would fill in these formulae as follows:

\[
\text{Prob}^{\text{TSS}} = \frac{e^{0.44}}{e^{0.44} + e^{-0.03} + e^{2.702}} = 0.09
\]

\[
\text{Prob}^{\text{PI}} = \frac{e^{-0.03}}{e^{0.44} + e^{-0.03} + e^{2.702}} = 0.06
\]

\[
\text{Prob}^{\text{GSD}} = \frac{e^{2.702}}{e^{0.44} + e^{-0.03} + e^{2.702}} = 0.85
\]

The sensory subtype with the highest probability is the subtype in which the child best fits. In the present example, the child is best characterized by the GSD subtype.