A sensory approach to the assessment and treatment of children with tic disorders

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BSc (OT)

A thesis submitted in the fulfilment of the requirements for the degree of

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CANDIDATE'S CERTIFICATE

I, *Nicolette Soler*, hereby declare that the work contained within this thesis is my own and has not been submitted to any other university or institution for any higher degree.

I, *Nicolette Soler*, hereby declare that I was the principal researcher of all work contained in this thesis, including work published by multiple authors.

I, *Nicolette Soler*, understand that if I am awarded a higher degree for my thesis titled, "S*ensory approach to assessment and treatment of children with tic disorders"* being submitted herewith for examination, the thesis will be lodged in the University Library and be will available immediately for use. I agree that the University Librarian (or, in the case of the department, the Head of the Department) may supply a photocopy or microform of the thesis to an individual for research or study or to a library.

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SUPERVISOR'S CERTIFICATE

This is to certify that the thesis entitled "*Sensory approach to assessment and treatment of children with tic disorders",* submitted by *Nicolette Soler* in fulfilment of the requirements for the degree of Doctor of Philosophy, is in a form ready for examination.

Professor Russell Dale

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DEDICATION

This thesis is dedicated to all children and young people with tic disorders and their families. Your courage, resilience and insight in dealing with the functional challenges resulting from tic disorders and comorbidities on a daily basis, are inspirational.

ABSTRACT

Tic disorders are the most common movement disorder in childhood, affecting ~1% of school-aged children. Tics are associated with multi-domain impairments, including social, physical and academic difficulties that interfere with the child's quality of life and functional performance in the home, school and community setting. With 80-90% of children with tic disorders experiencing comorbid neurodevelopmental disorders and/or neuropsychiatric conditions, assessing and treating children with tic disorders is complex.

In addition to the premonitory urge (sensation or discomfort before the tic), broader hypersensitivities have been reported in children with tics. Our patients frequently report these symptoms, yet there is limited research into sensory dysregulation symptoms in patients with tic disorders. Some patients have recounted using self-identified sensory-motor strategies to reduce their tics. With no available disease-modifying or curative therapies for tics and therapeutic interventions relying on alleviating symptoms, there is a clinical need for research into the prevalence of sensory dysregulation in children with tic disorders and to explore a sensory-based approach to managing tics.

Our first study aimed to understand the prevalence of sensory dysregulation symptoms in children with tic disorders (n=102) compared with age- and sex-matched healthy controls (n=61) through a cross-sectional study (Chapter 3). Sensory dysregulation, executive function, and quality of life data were obtained through the Short Sensory Profile-2 (SSP2), Child Sensory Profile-2 (CSP2), Sensory Processing Measure (SPM), Behaviour Rating Inventory of Executive Function-2, and Strength and Difficulties Questionnaire and Pediatric Quality of Life Inventory. Tic severity was assessed with the Yale Global Tic Severity Scale (YGTSS).

This study identified that children with tic disorders and comorbidities have significant sensory dysregulation symptoms compared to healthy controls ($P < 0.001$). There was a positive correlation between sensory dysregulation and global executive difficulties in children with tics and comorbidity ($n = 87$; $rho = 0.716$; $P < 0.001$) and a negative correlation between sensory dysregulation with quality of life ($n = 87$; $rho = –0.595$; $P < 0.001$). In children with tics, there was an association between sensory dysregulation and the number of comorbidities ($P < 0.001$). Thus, sensory dysregulation could be included in the neurodevelopmental symptoms of tic disorder.

The second study, an open-label prospective design, piloted a sensory-motor treatment approach with children with tic disorders (n=10) (Chapter 4). An adapted version of the Alert Program®, which used sensory-motor strategies to gain emotional regulation, was implemented with study participants over three 60–90-minute appointments. The YGTSS showed tic reduction in all participants, with a pre-intervention mean score of 46.5, improving to 17.7 post-therapy. These promising results warrant further research into sensory-motor intervention for tics to establish the effectiveness and feasibility of this approach. However, through these first two studies (prevalence (Chapter 3) and pilot studies (Chapter 4)), we identified that some sensory dysregulation symptoms were not identified using the SPM and CSP2. Therefore, this questioned the utility of these proxy-report sensory-based measures in children with tic disorders.

Given this uncertainty relating to these commonly used sensory measures with children with tic disorders, a systematic review was undertaken. This systematic review aimed to identify an appropriate measure with good psychometric properties to comprehensively assess sensory dysregulation symptoms of children with tic disorders (Chapter 5). From a systematic search

of 11 databases and hand searching by two reviewers, 7,352 articles were retrieved. Twelve proxy-report sensory-based measures for children and adolescents with neurodevelopmental disorders from 20 included articles were reviewed. The updated Consensus-based Standards for the selection of health Measurement Instruments (COSMIN) Risk of Bias checklist and criteria for good measurement properties to evaluate instrument development and psychometric properties were applied as the appraisal tools. Only one measure, the Participation and Sensory Environment Questionnaire-Home (PSEQ-H) Scale, had adequate content validity and met eight criteria for good measurement properties. Yet the items on any of these twelve measures did not include all sensory dysregulation experiences reported from patients with tic disorders. Therefore, there is a need to investigate the scope of the patients' reported sensory dysregulation experiences as the first step in developing a new comprehensive proxy-report sensory-based measure for children with tic disorders.

The fourth study uses semi-structured questions with 16 families to understand the breadth and impact of the lived experience of sensory dysregulation in children with tics from the perspective of the child and their parents (Chapter 6). The data from this qualitative study was analysed using thematic analysis. The findings confirmed that the sensory dysregulation experienced by children with tic disorders were broader than the scope currently being assessed on current proxy-report sensory-based measures, such as the CSP2, and SPM. In addition, a novel finding was that these sensory dysregulation experiences impact the entire family unit and their quality of life. Yet current sensory-based measures only evaluate the impact on the child with sensory dysregulation, not the family. This knowledge will aid in the item generation of the new sensory-based measure in our future research.

These four studies provide an extensive understanding of the prevalence of sensory dysregulation symptoms in children with tics, the breadth of these dysregulation experiences and the impact this has on daily life, which adds to knowledge. It is also evident that sensory dysregulation symptoms should be routinely assessed in children with tics disorders. Furthermore, the effectiveness of a sensory-motor approach to manage tic disorders was shown through the significant reduction in tics in the pilot study and warrants further research through a randomised controlled trial. Management guidelines for children with tic disorders should include recommendations relating to the inclusion of assessment and treatment of sensory dysregulation symptoms in children with tic disorders and comorbidities.

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DISSEMINATION OF RESEARCH

Parts of the research presented in this PhD thesis have been published (Chapter 3 (Study 1), Chapter 4 (Study 2) and Chapter 5 (Study 3)) or submitted to a journal for publication (Chapter 6 (Study 4)). The research has been presented in the following journals and conferences:

Peer-Reviewed Papers

- **Soler, N**., Hardwick, C., Perkes, I. E., Mohammad, S. S., Dossetor, D., Nunn, K., ... & Dale, R. C. (2019). Sensory dysregulation in tic disorders is associated with executive dysfunction and comorbidities. Movement Disorders, 34(12), 1901-1909.
- **Soler, N**., Hardwick, C., Perkes, I. E., Dossetor, D., Bray, P., & Dale, R. C. (2019). An exploratory study into an adapted use of the Alert Program® for tic disorder in children. Australasian Psychiatry, 27(2), 144-151
- **Soler, N,** Cordier, R, Perkes, IE, Dale, RC, Bray, P. Proxy-reported sensory measures for children and adolescents with neurodevelopmental disorders: A systematic review. Developmental Medicine Child Neurology. 2022; 00: 1– 15. https://doi.org/10.1111/dmcn.15367Published abstracts

Conference Presentations: Podium

- **Soler. N**, Hardwick. C, Perkes. I., Mohammad. S., Dossetor. D., Nunn, K., Bray. P, Dale R. Preliminary Investigation into sensory strategies for tic disorders in children. The University of Sydney Children's Hospital Westmead Clinical School Discipline of

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- **Soler. N**, Perkes. IE, Dale R. Bray. P. A systematic review of sensory assessment tools for children with neurodevelopmental disorders. 29th Occupational therapy Australia Conference, Sydney, Australia. 23-25th June 2021.

- **Soler. N**, Perkes. IE, Dale R. Bray. P. Research into Tourette's syndrome and comorbidities. Paediatric Acute-onset Neuropsychiatric Syndrome, (PANS) Multidisciplinary Approaches Conference, Sydney, Australia. The 5th of June 2021.
- **Soler. N**, Perkes. IE, Bray. P, Dale RC. Three Minute oral thesis: Improving the future of children with tic disorders through a novel multi-modal approach. World Federation of Occupational Therapy (WFOT). Paris, France. 28-31st of August 2022.
- **Soler. N**, Perkes. IE, Bray. P, Dale RC. A systematic review of sensory assessment questionnaires used for children with neurodevelopmental disorders. World Federation of Occupational Therapy (WFOT). Paris, France. 28-31st of August 2022.

Conference Presentations: Oral Poster

- **Soler. N**, Hardwick. C, Perkes. I., Mohammad. S., Dossetor. D., Nunn, K., Bray. P, Dale RC. An exploratory study into the use of a sensorimotor approach for tic disorder in children. 10th European Society for the study of Tourette's syndrome Conference (ESSTS). Seville, Spain 14-16th June 2017.
- **Soler. N**, Hardwick. C, Perkes. I., Mohammad. S., Dossetor. D., Nunn, K., Bray. P, Dale R. Prevalence of sensory and emotional features in children with tic disorders. Hospital Week Research Symposium. The Children's Hospital at Westmead. The 31st of August 2018.
- **Soler. N**, Hardwick. C, Perkes. I., Mohammad. S., Dossetor. D., Nunn, K., Bray. P, Dale RC. Prevalence of sensory symptoms and emotional regulation in Australian children with tic disorders. European Society for the study of Tourette's syndrome Conference (ESSTS). Copenhagen, Denmark. June 2019.

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Additional Work During Candidature Not Forming Part of the Thesis

- Jones, H. F., Han, V. X., Patel, S., Gloss, B. S., **Soler, N**., Ho, A., ... & Dale, R. C. (2021). Maternal autoimmunity and inflammation are associated with childhood tics and obsessive-compulsive disorder: Transcriptomic data show common enriched innate immune pathways. *Brain, Behavior, and Immunity*, *94*, 308-317

Thesis Outline and Structure

This section outlines the stages of research undertaken at a large tertiary paediatric tic disorder clinic in Sydney, Australia, to examine the influence of a sensory-based approach in reducing tic severity and intensity. There are four studies that together explain the learning undertaking in terms of assessment and proposed treatment with a sensory-motor approach to managing tic disorders in children.

The thesis includes traditional chapters (chapters one, two and seven), three peer-reviewed papers (chapters three, four and five) in published format and chapter six in the structure of a journal article as the manuscript has been submitted for publication to a journal. Chapters three through six form individual chapters and explain each of the four studies conducted as part of this thesis.

This PhD thesis contains seven chapters:

Chapter one *(this chapter)* provides background and context to the research studies and thesis, explains the research need, defines the research scope, and outlines the research aims. The problems to be addressed are presented while stating the directions, aims and objectives of the thesis.

Chapter Two reviews the literature on the clinical profile of tic disorders and sensory dysregulation symptoms in children and adolescents with tic disorders and other existing neurodevelopmental disorders.

Chapter Three provides details and results of a cross-sectional study of 163 children (n=102 children with tic disorders and n=61 neurotypical controls) to investigate the prevalence of sensory dysregulation in children with tic disorders and comorbidities.

Chapter Four describes the findings from a study piloting a novel modified sensory-based approach to treating tics with 12 study participants with a primary tic disorder. The treatment approach adapted the Alert Program®, designed to use sensory-motor strategies for emotional regulation as a method to reduce tic severity and intensity. This approach has been used successfully with other neurodevelopmental disorders but not trialled with children with a tic disorder.

Chapter Five reports the findings of a systematic review of proxy-reported sensory-based measures for children and adolescents with neurodevelopmental disorders. Following the systematic search of 11 databases, data were extracted from 20 articles relating to the 12 identified measures. COSMIN was used as the appraisal tool to evaluate content validity and psychometric measures. All psychometric properties of all 12 sensory-based, proxy-reported measures are presented to facilitate evidence-informed clinical decision-making.

Chapter Six presents a qualitative study of 16 families with children or adolescents with tic disorders to understand the breadth and functional impairment of sensory dysregulation experiences in their daily lives. The information gained relating to sensory dysregulation symptoms from families in this final study is the first step in co-designing a comprehensive, comprehendible, and relevant proxy-reported sensory measure with patients in the future.

Chapter Seven discusses the results and limitations of the thesis, explores the implications of the thesis for clinical and research practice, and identifies future research directions.

CHAPTER ONE

Introduction

This chapter aims to provide an understanding of the context, need for, and relevance of the research into sensory dysregulation in children with tic disorders. The research need, the aims of the research, and a summary of the thesis outline and structure are provided.

1.1 Research Terms

Tic Disorders

Tic disorders are the most common movement disorder in childhood (1). Children with tics suffer from repetitive, stereotypical, rapid, non-rhythmic movements or vocalizations (2). Tic disorders are classified as neurodevelopmental disorders and typically present during the prepubertal period, with the average onset period between four and six years of age (2). The peak severity of the tics is experienced between the ages of 10 and 12 years, followed by a reduction of tic severity in adolescence and adulthood (3).

Premonitory Urge

The phenomenological description of tic disorders includes a sensory phenomenon, such as a feeling of tightness, tension or itching experienced before the tic expression, known as the premonitory urge (3, 4).

Sensory Dysregulation

The ability to adaptively organise and regulate responses to sensory stimuli (these include hearing, vision, touch, smell, taste, movement and balance (vestibular), body awareness (proprioception) and interoception in one's environment is critical to participation in everyday activities (5). The nature of the responses to this sensory input from either our body or our environment is expected to be appropriate, graded and adaptive so that the person can maintain an optimal range of performance and adapt to challenges (5). Atypical responses to sensory stimuli can result in behaviours incongruent with the sensation experienced (6). For instance, a person may over- or under-respond in their responses to sensory input in a manner disproportionate to the sensation experienced (5). For example, as frequently reported by our patients, a child may be unable to attend school due to the intolerable discomfort and distress

associated with the feeling of the material, tags and seams associated with school uniform on their skin.

The sensory features of neurodevelopmental disorders are variously described as ‘‘sensory dysregulation’’, ‘’sensory regulation’’, ‘‘sensory phenomena’’, ‘‘sensory modulation disorder’’, ‘‘sensory processing’’, ‘‘sensory reactivity’’ and ‘‘atypical sensory reactivity’’ (2, 5, 7-9). At the commencement of this PhD, we had many discussions within the multi-disciplinary supervision team relating to the correct terminology to describe these sensory features. In this thesis, the term ‘sensory dysregulation’ will be used (5, 7, 9, 10) except in chapter 4 (study 2). Study 2 (Chapter 4) was the first of our studies to be published, and the term ‘‘sensory sensitivities’’ was used rather than ‘‘sensory dysregulation’’ as, at the time, this was determined to be the more accurate terminology. As the research has continued, we have gained a better understanding of the terminology used and, as a multi-disciplinary research team, have come to the consensus of the term ‘sensory dysregulation’.

Functional Impairment

This thesis discusses the impacts on a person’s ability to function due to tic disorders or sensory dysregulation symptoms. The International Classification of Functioning and Health (ICF) definition of ‘‘functioning’’ states that this is an all-encompassing term that extends to body functions, body structures, activities and participation (11). It is understood that interactions between the individual and their environment and personal factors enable this functioning to occur (11). For this thesis, the difficulties that a person or patient is experiencing will be described as functional impairment.

1.2 Research Background

Tic disorders are the most common movement disorders in childhood, affecting 1 in 100 children and adolescents (4, 12, 13). Children with tics may experience pain, bullying, social isolation and reduced self-esteem. Downstream effects include reduced school attendance, poor learning and academic performance, and difficulty with independence in self-care tasks (i.e. feeding oneself) (1, 4). ‘‘Sensory phenomena’’ is often described by people experiencing tics and include the premonitory urge and ‘‘just right’’ perceptions (14). Eighty to ninety percent of children with tic disorders are diagnosed with other neurodevelopmental and neuropsychiatric conditions such as obsessive-compulsive disorder (OCD), attention deficit hyperactivity disorder (ADHD), autism spectrum disorder (ASD) and depressive disorders (1, 2, 4).

Treatment of tic disorders needs to be driven by the effects on the child’s quality of life and the functional impairment resulting from tics. Factors that influence care planning for optimal management and treatment for the individual child consider i) the severity of the tics, ii) the effect of the tic disorder on daily function, iii) the impact on the child’s quality of life, and iv) determining the most debilitating symptoms, bearing in mind these symptoms may pertain to comorbidities or co-existing conditions rather than the tic disorder *per se* (4, 13).

Tic disorders are treated with a judicious combination of psychoeducation, behavioural and pharmacological approaches (4, 13). Providing psychoeducation to the child with tics, their parents, teachers, and peers is an essential first step of treatment (4, 13). Behavioural therapies are considered a second-line treatment for tics, and pharmacological treatments are reserved for severe cases (4).

Although non-pharmacological treatment approaches, such as Comprehensive Behavioral Intervention for Tics (CBiT), have been proven effective in reducing tics (15), there are limitations to these psychological approaches. The mechanisms by which behavioural therapy is effective are unclear (4). However, children achieve modest therapeutic gains with CBiT, with a major confounder being the intensity and considerable effort required due to the program being time- and resource-intensive (15). Children under ten years of age may not be aware of the premonitory urge or be able to understand and apply the treatment strategies of CBiT (13). Therefore, current behavioural therapies such as CBiT are less feasible in young children (under ten years of age) or where comorbidities are present, e.g., ADHD, intellectual disability, and ASD (13).

Pharmacological treatment is typically reserved for severe cases, such as when tics are causing pain or injury, social and emotional issues and/or functional impairment (1, 4, 13). Although pharmacological treatment effectively reduces tic frequency and intensity, treatment only dampens the frequency and intensity of the tics and may produce side effects (1, 4, 13). Therefore, it is common practice to postpone pharmacological treatment of tics until impairment justifies the risk-benefit paradigm (16). Invasive therapies such as botulinum toxin or deep brain stimulation are only considered in refractory and severe cases (1, 13, 16, 17). Thus it is evident that there is a need to find alternative, complementary treatment methods for children experiencing tic disorders where existing treatment is inadequate.

A paediatric tic clinic was established at a large tertiary children's hospital in Sydney, Australia, with a multi-disciplinary team comprised of a paediatric neurologist, psychiatrist, psychologist and occupational therapist. This clinic is unique as an occupational therapist is included in the multi-disciplinary team, and a sensory-based approach is being investigated to

complement CBiT, or trial where CBiT is not recommended to manage tics. Occupational therapists are predominately included in the multi-disciplinary team providing assessment and treatment to children with neurodevelopmental disorders (18-23). However, even though tic disorders are also classified as a neurodevelopmental disorder (2), it is not standard practice for occupational therapists to be part of the multi-disciplinary team providing care regarding tic management. By contrast, sensory-based treatment approaches are provided by occupational therapists to children with neurodevelopmental disorders, such as ASD and ADHD (18, 23, 24). However, this is not the case for children with tic disorders. In rare instances when occupational therapists are involved in treating children with tic disorders to manage tics and not the comorbidities, CBiT is provided and not a sensory-based approach (25).

Through the clinical experience of working in this multi-disciplinary tic disorder clinic as an occupational therapist, it was observed that children with tics reported sensory dysregulation symptoms and identified their own sensory strategies, i.e., deep pressure gained from wearing tight gloves was explained by a patient to reduce his finger-tapping tic. When reviewing the literature, only a single study investigated sensory dysregulation in children with tic disorders (25). This Canadian study by an occupational therapist reported children with tic disorders and ADHD experienced significant sensory dysregulation symptoms (25). This study highlighted several gaps in the current research concerning assessment and treatment in children with tic disorders.

This led my colleagues and I, the clinicians in the Sydney tic clinic, to examine the prevalence of sensory dysregulation symptoms in children with tic disorders and other comorbidities seen through this tertiary paediatric service. As there were no guidelines to specify the appropriate

sensory-based assessment measure to be used with children with tic disorders, we sought to identify valid, reliable and fit-for-purpose patient/proxy-report sensory-based measures for use with children with tic disorders so as to be used in our clinic and research studies. Finally, we aimed to understand whether a sensory-based treatment approach to manage tic disorders would be effective, as used with children with other neurodevelopmental disorders.

1.3 Research Need

With tic disorders being the most prevalent movement disorder and no disease-specific treatment, there is a critical need for a feasible complementary assessment and treatment program for children with tic disorders (1, 4).

With 80-90% of children with tic disorders having co-existing neuro-developmental or neuro-psychiatrist comorbidities (1, 4, 13), and a paucity of existing literature (25), there was a need to understand the prevalence of sensory dysregulation symptoms in children with tic disorders (and associated co-morbidity). As only a single study had researched the sensory dysregulation symptoms in children with tic disorders using the Short Sensory Profile (25), the question arose whether other proxy-report sensory-based measures, such as the Sensory Processing Measure (26), would be suitable for use in his population. Therefore, there was a need to examine which of the commonly used proxy-report sensory-based measures, the Sensory Profile 2 or Sensory Processing Measure had better utility in assessing sensory dysregulation in children with tic disorders.

Furthermore, a new treatment approach is needed to complement existing treatment for children with tics. In particular, i) for children with tics younger than ten years, ii) for patients where CBiT is not feasible, iii) where the tics are not severe enough for medication use but additional treatment is still required, or iv) where there is complexity due to comorbidities (13,

27). With the emergence of evidence of sensory dysregulation symptoms associated with tic disorders in addition to the 'sensory phenomena' (e.g. premonitory urge and 'just-right' perceptions), then novel approaches such as those that target sensory symptoms may be useful in children with tic disorders (1, 25).

One such sensory-motor treatment approach used effectively with children with other neurodevelopmental disorders is the Alert Program® (28). The Alert Program® uses sensory-motor strategies to assist children and adults with self-regulation and attention skills (29). Although the sensory-motor-based treatment approach, such as the Alert Program®, has been implemented with children with other neurodevelopmental conditions, it has not previously been trialled with children with tic disorders. As it has been reported that emotional dysregulation increases tics, (12) using a sensory-motor approach such as the Alert Program®, which addresses emotional dysregulation through sensory-motor strategies, may reduce tic frequency and intensity.

1.4 Aims of the Thesis

This thesis aimed to examine a novel assessment and treatment approach for children with tic disorders. In doing so, the researchers determined the i) prevalence of sensory dysregulation symptoms in children with a tic disorder and comorbidities, ii) piloted a novel treatment approach using sensory motor strategies to manage tic disorders, iii) identified and evaluated the current proxy-reported sensory-based measures and iv) worked with young people and their parents to begin the co-design of a new sensory measure.

The following objectives were developed for the thesis:

1. To measure the prevalence of sensory dysregulation symptoms in tic disorders and their clinical associations in a quaternary clinic sample.
2. To pilot test the effectiveness of a sensory motor-based therapy to treat tic disorders in children.
3. To determine the quality and utility of proxy-reported sensory-based measures for children and adolescents with neurodevelopmental disorders.
4. To understand the breadth of sensory dysregulation and functional impairment in daily life as experienced by young people with tic disorders and their parents.

CHAPTER TWO

Literature Review

The purpose of this chapter is to critically review the literature to provide an overview and understanding of tic disorders and sensory dysregulation in children with tics. As tic disorders are classified as neurodevelopmental disorders in the Diagnostic and Statical Manual of Mental Disorders (DSM-V), a broad context of neurodevelopmental disorders is first provided. This chapter is organised into two sections, with section 2.1 focusing on tic disorders and section 2.2 addressing sensory dysregulation. Both sections will explain the conditions, functional impairments, assessment measures, and current treatment practices.

2.1 Overview of Tic Disorders

2.1.1 A Broad Context of Neurodevelopmental Disorders

Tic disorders are conceptualised as neurodevelopmental disorders (2). Neurodevelopmental disorders are a heterogeneous group of conditions with an onset during the developmental period (2). These disorders are characterised by neurodevelopmental deficits and delayed milestones, causing impairment(s). Neurodevelopmental conditions include attention-deficit/hyperactivity disorder (ADHD /ADD), autism spectrum disorder (ASD), developmental coordination disorder (DCD), intellectual and global developmental delays (ID and GD), specific learning disorders, communication disorders and motor disorders (2). Motor disorders include tic disorders and Tourette's syndrome, and stereotypic movement disorders (2). Symptoms and impairment in functioning relating to neurodevelopmental disorders commonly manifest before children commence primary school, but children's age at the time of diagnosis is variable (2). Prevalence rates of neurodevelopmental disorders are between 10 to 15% of all births, although this prevalence is understood to be increasing (30).

Neurodevelopmental disorders can cause limitations to one or multiple areas of daily functioning, including attention, executive function, intelligence, learning, language, motor skills, sensorimotor deficits, social relationships and behaviour (30). These deficits in skills cause impaired personal, social, academic and/or occupational function (2). The presentation of symptoms exists along a continuum of severity, and the impairments and behaviours can change as the child develops and matures (30). The range and severity of impairment(s) vary and are further complicated by the possible presence of multiple co-existing neurodevelopmental and neuropsychiatric disorders (2).

2.1.2 Tic Disorders and Tourette's Syndrome in Childhood

Tic disorders are the most common movement disorder in childhood (1). The peak severity of tics is experienced between the ages of 10 and 12 years, typically followed by a reduction of tic severity in adolescence and adulthood (3). One-third of children diagnosed with Tourette's syndrome (TS) will be free of tics into adulthood (4). Less than 5% of individuals report experiencing a worsening of tics into adulthood (4).

As with other neurodevelopmental disorders, tic disorders are more common in males than females, with the ratio varying between 2:1 to 4:1 (3). Tic disorders and TS affect all races and ethnicity. Tic disorders do not differ in clinical characteristics, course or etiology according to race or ethnicity, but culture may impact how tic disorders are perceived and managed (1).

Tics are commonly experienced in a waxing and waning course (4). The specific type of tic disorder is defined through the clinical presentation and course of the condition (2). There are four diagnostic categories for tics as per the Diagnostic and Statistical Manual of Mental Disorders 5th edition (DSM-V): i) Tourette's syndrome, ii) persistent (chronic) motor or vocal tic disorder, iii) a provisional tic disorder, and iv) other specified and unspecified tic disorders (2). When a child or adolescent has experienced both motor and vocal tics in a waxing and waning pattern for one year or more, the condition is classified as Tourette's syndrome or, more accurately, Gilles de la Tourette syndrome (2). Should the child only experience persistent vocal or motor tics, the condition is classified as chronic vocal or motor tic disorder, respectively. When the tic condition has been epresent for a period of less than one-year period, the tic disorder is referred to as a provisional tic disorder (2).

2.1.3 Pathophysiology, Genetics and Environmental Factors of Tic Disorders

Abnormalities in pathways between the cerebral cortex and basal ganglia, resulting in neuronal dysfunction of both motor and limbic systems, are hypothesised to represent the most likely pathophysiology of tics (13, 31). The sensory limbic and executive corticostriatal loops are also impacted (in addition to the motor circuits), resulting in impairment in these functions (13, 31, 32).

Genetic and environmental factors (including in-utero and ex-utero factors) influence the phenotypic expression of the disorder (2, 13). Tic disorders have been demonstrated to be one of the most heritable, non-Mendelian neurodevelopmental disorders, with first-degree family members 10-100 times more likely to be diagnosed with the condition than the general population (13). Associated risk genes for TS of significant effect have been identified (4, 12). Although several chromosomal regions of interest have been identified through linkage studies, only histidine decarboxylases (HDC) and neurexin 1 genes have been associated with a rare coding mutation in more than one study (therefore reproduced) (1). It is important to note these rare genetic variants only account for a small proportion of patients with TS(1). Low levels of histamine, a key basal ganglia neurotransmitter, in the basal ganglia have, in recent studies, been suggested to cause the symptoms associated with TS (33).

Environmental factors such as low socioeconomic status and maternal illness during pregnancy, which include depression and autoimmune diseases, as well as complications during pregnancy and birth, also appear to play a role in tic expression in the child (2, 34, 35). Factors such as stress, anxiety, excitement and fatigue exacerbate tics in the affected individual

(12). As tics are suggestible, when associating with other people with tic disorders, new tics can appear in the person's repertoire of tics (12).

Although tics can include almost any muscle group, specific movements and vocalisations are common across patient populations, such as eye blinking, sniffing, or throat clearing (2). Most motor tics are 'above the shoulders', meaning the tics are experienced in the areas of the shoulders, head and face. However, the involvement of the trunk and limbs does also occur, often in more severely affected patients (1) (Refer to Chapter 2, Table 1 for common motor and vocal tics). Tics can either be simple motor or vocal tics that are short, lasting milliseconds (i.e., eye blink, shoulder shrug, sniff or a grunt) or complex tics, which are less common, lasting seconds. Complex tics often involve a combination of simple tics performed in a specific pattern (i.e., head turn and shoulder shrug), known as an 'orchestra' of tics (1, 4). Vocal or motor tics change over time and are replaced by new movements and/or vocalisations (13). Tics are understood to have voluntary and involuntary aspects, as there is a 'suppressible' aspect to tics, with many individuals able to suppress their tics for a limited period, causing mounting discomfort (1, 4).

Table 1: List of the most common simple motor and vocal tics (36).

Common Motor Tics	Present %	Common Vocal Tics	Present %
Eye blink	72%	Sniffing	42%
Head jerk	67%	Grunting	31%
Arm/ hand movements	61%	Other vocal tics	29%
Shoulder shrugs	56%	Coughing	23%
Facial grimace	55%	Blocking/stuttering	19%
Mouth/ tongue movements	55%	Words	15%
Leg/feet movements	50%	Echolalia (repeating vocalisations of others)	12%
Chest/stomach tightening	37%	Snorting	12%
Other motor tics	32%	Phrases	11%
Eye rolling/darting	31%	Syllables	10%
Pelvic tensing movements	17%	Animal noises	8%
Echopraxia (copying another's gestures)	8%	Coprolalia (obscene words)	7%
Copropraxia (obscene gestures)	4%	NA	NA

The percentage of prevalence and list of common motor and vocal tics are from an American cohort of n=122 study participants (36).

Tourette's syndrome is associated with echophenomenon, palliphenomenon and coprophenomenon (37). Echophenomenon is a term that incorporates echolalia (the imitation of sounds or sounds) and echopraxia (the imitation of actions) and presents in approximately 11 to 44% of patients with Tourette's syndrome (37). A substantial portion of patients with Tourette's syndrome will experience palliphenomenon, which is repeating behaviours or actions such as saying a word repeatedly (palilalia) and forced touching of objects or body parts (palipraxia) (37). Although rare, Tourette's syndrome is sometimes associated with coprohenomena, which is the involuntary expression of socially inappropriate words or gestures (37, 38). Coprolalia (swearing tics) is experienced by 7-20% of individuals with Tourette's syndrome and typically presents five years after the initial onset of the disorder (13, 36-38). Coprolalia is three times more commonly encountered than copropraxia (socially unacceptable motor tic, such as rude hand gestures) and is observed in 6 % of males and 5 % of females (38). Tic severity and the presence of coprohenemona are synonymous with considerable social stigma and poor quality of life (2). Patients with pure tic disorders have been shown to have a better quality of life than those with comorbidities, although 80-90% of children with tic disorders have co-existing comorbidities (4).

2.1.4 Premonitory Urge and Just-Right Experiences

The premonitory urge is described as localised discomfort immediately before a tic and may contribute to tic generation (39). The sensation is aversive or unpleasant; in some cases, our patients have described this experience as worse than the actual tic. This sensation builds up

until the tic is completed, followed by a relief of these sensory 'urge' symptoms (3). The prevalence of the premonitory urge varies between studies, from 8.5% to 96% (3). Higher rates of premonitory urge are associated with an increase in age (i.e., 24% in children aged 8 to 10 years, 34% in children 11 to 14 years and 57% in adolescents between 15 and 19 years) (3). Although the typical age of onset for the premonitory urge is ten, and generally three years following the onset of the tic disorder, it is believed the premonitory urge may always be present (1, 3). It is thought that the child's awareness of the premonitory urge from age ten is a result of greater cognitive awareness with maturity (1).

2.1.5 Complexity of Comorbid Neurodevelopmental Disorders and Tic Disorders

The majority (80-90%) of children with tic disorders are afflicted by other neurodevelopmental and neuropsychiatric conditions such as OCD (30-40%), obsessive-compulsive behaviours (OCB) (60-90%), ADHD (54-60%), ASD (5-15%) and depressive disorders (13-76%) (4, 13, 40, 41). The presentation of OCD associated with tics relates more to symmetry, counting, and touching (13). In contrast, OCD in the absence of tics tends to be more associated with contamination obsessions and compulsions (13). Anxiety, externalising disorders (such as oppositional defiant and conduct disorders), learning disorders, sleep disorders and impaired social cognition difficulties are also common in people with tics and TS (13). In addition, sensory dysregulation has been noted in children with tics and ADHD (25).

Tic disorders and TS are complex conditions, particularly when associated with multiple comorbidities, which negatively impact the quality of life for children and adolescents. The motor and vocal tics are the observable symptoms covering the 'tip of the iceberg', with many comorbidities below the surface (Chapter 2, Figure 1). As many as 27% of patients may have three or more comorbidities in the presence of a tic disorder (1, 4). With patients with multiple

comorbidities and tics, the most debilitating comorbidities can change at different time points (1). This concept may be explained to patients using the analogy of a block of appartments when describing this relationship. Each apartment represents different comorbidities patients may have in addition to the tic disorder. The most debilitating condition at any given time resides in the penthouse, and the condition with the least impact is placed in the basement. The placement of these conditions concerning the severity and functional impairment can change (1). For instance, tics wax and wane; therefore, in a waxing phase, this tic disorder could be the most debilitating of all the conditions and comorbidities the patient experiences (1, 4). Thus, for children and families to manage these complexities, each patient requires an individualised treatment plan (4).

Figure 1: Tourette's syndrome: Tics are just the tip of the iceberg, with multiple comorbidities below the surface, demonstrating the complexity of tic disorders. This illustration is from the Tourette Association of America website 2021 resource library.

2.1.6 Functional Impairment in Children with Tic Disorders

Children and adolescents with chronic tic conditions often experience mild to moderate functional impairment (42). The degree of functional impairment usually depends on tic severity and co-occurring comorbidities (1). The comorbidities often predict the degree of functional impairment more than the tic disorder itself (1). These functional impairments are associated with multi-domains, including social, physical and academic difficulties that interfere with the child's quality of life and functional performance in the home, school and community setting (43). Children with tics experience discrimination, bullying, social isolation and reduced self-esteem, which correlates with loneliness, anxiety and internalising symptoms (42, 43).

Neuropathic pain or tissue damage resulting from the frequency and intensity of tic movements can be experienced as a physical impact (43). Headaches, neck and shoulder pain, and even stress fractures can result from tics (44, 45). In rare but severe cases, the force of the tic can cause injury, such as herniation of a cervical disc due to a neck tic (13).

Children diagnosed with tic disorders experience adverse functional impairments in learning and academic performance due to the secondary effects of tics (42). Performing well on time-limited tasks, handwriting, answering questions, reading aloud and completing homework are all tasks that can be negatively affected by vocal / and or motor tics (42). Difficulties with attention and concentration may be from the disturbance of the tics and/or co-existing attention deficit disorders (42). Stigma and social maladjustment experienced by children with tics in the school setting are commonly the results of a misunderstanding of the disorder by peers and teachers (46). Parents, caregivers and teachers frequently interpret tics as rude behaviour, such as an eye roll or shoulder shrug, and the child may be punished (46). The stress of being

misunderstood, unfairly treated and punished can create stress, and this emotional distress may increase the frequency and intensity of the tics displayed (46). A Swedish study conducted with 7736 people with tic disorders over 44 years determined that tic disorders are associated with a substantial risk of suicide (47).

Parents of children with tic disorders experience high caregiver strain and stress, and parents have a higher risk of psychiatric morbidity (48). The daily challenges experienced by parents and caregivers of children with tic disorders include i) managing their child's challenging behaviours, ii) misconceptions and lack of understanding of the condition by lay people and health professionals, iii) negative experiences related to their child's education and iv) a lack of support and services for families with a child with tic disorders (49). Downstream effects include reduced school attendance, poor learning and academic performance, and difficulty with independence in self-care tasks (i.e., feeding oneself) (43).

2.1.7 Assessment Measures to Evaluate Tic Disorders in Children

Both clinician and parent-reported assessment measures are used to evaluate the severity, intensity and functional impairment of tics. The Yale Global Tic Severity Scale (YGTSS) is the gold standard clinician-rated measure for tic-based assessments with acceptable psychometric quality (50-52). The use of the YGTSS is recommended in the European Child Adolescent Psychiatry guidelines for evaluating tic disorders (50).

The YGTSS is a semi-structured clinical interview that takes approximately ten minutes to complete and is used with children aged six to seventeen years (13). It provides a multidimensional assessment of tic symptom severity across five dimensions: i) number, ii) frequency, iii) intensity, iv) complexity, and v) interference of both motor and phonic tics over

the past 7–10 days (50-52). Each domain (for both motor and vocal tics) is scored separately on a six-point ordinal scale (0-5) anchored with descriptive statements and relevant examples (52). Clinicians are provided with five summed scores to interpret the severity of their patient's tics. The scores for the motor tics are added to give a total motor tic score (range 0-25) and, likewise, a total vocal tic score (range 0-25) (50). The total motor and vocal tic scores are combined to produce a total tic score (range 0-50). This total tic score is then added to the overall impairment score (range 0-50) to provide the clinician with the Global severity score (range 0-100) (50). The higher the score, the greater the impairment experienced. The YGTSS has been the most commonly used tool in clinical trials and clinical practice since it was developed in 1989 (50).

The Parent Tic Questionnaire (PTQ) is a proxy report measure completed by parent/caregiver designed to assess the presence, frequency and intensity of both motor and vocal tics in children and adolescents during the previous week (53). The PTQ follows the format of the YGTSS and provides a list of fourteen common motor and fourteen common vocal tics. Parents are asked to score the frequency and intensity of each of these twenty-eight listed tics (53). The frequency (rated: weekly, daily, hourly, or constantly), intensity (rated: mild, noticeable by others and the forcefulness of the tic), and the pain experienced as a result of the tics are rated on a four-point scale (range 1 - 4) (53). A motor and vocal tic score is provided, as is a total severity score (53). The PTQ measure was demonstrated to have excellent internal consistency and good to excellent test-retest reliability (53).

The Premonitory Urge for Tics Scale (PUTS) is a measure that rates the pre-tic urges (premonitory urge) as well as the relief that may be experienced after the tic has been completed (3). The person with a tic disorder rates each of the nine statements relating to their experience of the PU as either i) not at all true, ii) a little true, iii) pretty much true, or iv) very much true

(3). The measure uses a four-point ordinal scale to score each of the nine items (3). A total score is calculated by summing all nine items. Thus, total scores range from nine to thirty-six, with higher scores indicative of greater premonitory urge symptoms (3). The PUTS questionnaire was determined to have good internal reliability in children and adolescents, but due to the age-related difference reported when children can identify the PU, there is a need to develop an age-specific questionnaire to asses urges (54). Through exploratory factor analysis, the PUTS distinguished between sensory phenomena related to tics and mental phenomena found in OCD in children 11 years and older (54). Although the PUTS provides information on sensory phenomena, it does not assess sensory dysregulation symptoms.

It is not standard practice or part of clinical assessment guidelines to include the evaluation of sensory dysregulation as part of the holistic assessment of children with tic disorders (55). As recently as 2020, a new self-report questionnaire for tic-association sensations was developed, the RASTS (Rumination and Awareness Scale for tic -associated sensations) (54). In addition to the tic-associated sensations measured by the PUTS, the RASTS questionnaire assesses two additional aspects of tic-associated sensations in children (older than ten years) and adults (56). These include the intensity of somatosensory hyperawareness and the ability to identify signals of emerging tics (56). The ‘‘somatosensory hyperawareness’’ being measured in this questionnaire relates to the sensations of the PU experienced. It is not hypersensitive to other sensory experiences the person with tics may encounter from their body or environment (56). Nonetheless, this and a previous study by Jewers et al. (2013) (25) show growing interest in sensory dysregulation in people with tics and a need to consider the inclusion of sensory dysregulation symptoms when assessing the holistic functional impairment of children with tic disorders.

In addition to these above-mentioned tic-related measures, the Practice Guideline Recommendation Summary of Treatment of Tics in People with TS and Chronic Tic Disorders outlines the inclusion of assessment of comorbidities such as ADHD, OCD and other psychiatric comorbidities in children with tics (55).

2.1.8 Therapeutic Approaches for Tic Disorders in Children

Treatment of tic disorders is driven by the impact on the child or adolescents' quality of life and the functional impairments they experience (1, 4). Tic disorders are treated through psychoeducation, psychological and pharmacological approaches or a combination of these (4, 13). Although evidence supports the implementation of various behavioural, pharmacological, and neurostimulation treatment approaches for tic reduction, the benefits and harm associated with the various interventions must be considered (57). Providing psychoeducation to the child with tics, their parents, teachers, and peers is an essential first step of treatment (4, 13). Identifying associated coping strategies is typically sufficient in managing children experiencing mild to moderate tics (4). Pharmacological or psychological intervention of tics is unnecessary in many patients with less severe tics (1). Psychological therapies using a behavioural approach are considered before pharmacological treatments in the treatment for tics (4).

In terms of non-pharmacological treatment, behavioural therapies such as Habit Reversal Therapy (HRT), and Exposure and Response Prevention (ERP) are the most evidence-based behavioural interventions for tics (4, 13). HRT is comprised of three primary components: i) awareness training (aimed at noticing the PU or tic onset), ii) competing response training (an action that is incompatible with the target tic movement is taught), and iii) social support (praise

of the competing response being implemented) (4). Weekly treatment is provided to treat a single tic at a time through HRT (4).

CBiT is a treatment approach encompassing HRT, function-based interventions, relaxation training, psychoeducation and a reward-based procedure aimed at treatment compliance (4). CBiT has been proven effective in reducing tics (15). The treatment can be delivered via telehealth while reporting treatment gains (58). For children between the ages of 9 and 17 years of age with tics, CBiT is the preferred treatment over supportive psychotherapy (13, 27). CBiT was found to have high confidence over psychoeducation and supportive therapy to reduce tics (57). Patients who have responded well to CBiT have maintained these treatment gains for at least six months (55). Practice guidelines for the treatment of people with TS and chronic tic disorders recommend that people with tics who have access to CBiT, should be offered CBiT as an initial form of treatment (55). Evidence demonstrates that CBiT offers no increased risk of adverse effects for people with tic disorders compared with those treated with psychoeducation plus supportive therapy (55). Research has demonstrated that the effect size for CBIT appears similar to the effect sizes for pharmacological intervention (55).

ERP focuses on the patient actively suppressing tics during treatment sessions to increase their tolerance to the premonitory urge (4, 13). All the tics are treated simultaneously using the ERP approach, unlike CBiT, which addresses a single tic at a time (4).

The mechanisms by which behavioural therapy is effective are unclear (4). CBiT trials conducted with children and adults demonstrated the effectiveness of an 8-session protocol; however, patients with poor tic awareness, reduced treatment motivation, more severe tics, or substantial clinical comorbidity require a longer course of therapy (55). Barriers to

implementing behavioural therapy as a tic treatment include the cost and time-intensive nature of the program and the ability to access trained professionals experienced in implementing the program (15). For many patients with a tic disorder, access to CBiT is extremely limited due to financial restraints and a lack of trained health professionals. The willingness of patients to engage can serve as a further barrier (4, 55). Current therapies such as CBiT are more challenging in young children (under ten years of age) and where comorbidities are present (e.g., ADHD, intellectual disability, ASD) (55). Children under the age of ten may not be aware of the premonitory urge or be able to understand and apply the treatment strategies (13, 55). Although CBiT has been implemented in children aged nine years or younger, there is little evidence to demonstrate the effectiveness of this intervention in this age range (55).

Further research into psychological interventions for tics is required (55). There is a need to understand the efficacy of CBiT compared with pharmacological treatments for tics (55). Additional research is also necessary to determine the effectiveness of CBiT and HRT delivered via teleconferencing compared to face-to-face therapy appointments (55). In terms of HRT, further research is required to determine the relative efficacy of HRT compared with exposure and response prevention or educational group treatment in reducing tic severity (55).

The aim of using a pharmacological treatment approach for tic disorders is to improve the tic disorder and assist with psychosocial function (4). The use of pharmacological treatment is based on the theory that there is a neurochemical imbalance in tic disorders, and the medication targets dopaminergic, adrenergic, or other neurochemical pathways that can benefit the patient's symptoms (1). Anti-tic medication is reported to reduce tics by 25-70%, dependent on dose, over a two to four-week period (4). Pharmacological treatment, used only in severe cases, is considered when tics are causing pain or injury, social and emotional issues and/or

functional impairment (4, 55, 57, 59). There is no evidence that implementing pharmacological treatments for tics alters the long-term clinical course of TS (12). The gaining of consensus in a psychopharmacological treatment approach has been impeded due to the waxing and waning nature of the course of tics and the presence of comorbid and co-existing conditions (4). Pharmacology is often determined not just by the tic severity but by the co-existing comorbidity. Standardisation of a pharmacological approach to tics is problematic because the required doses, response time and efficacy are highly variable (4).

Pharmacological treatment only dampens the frequency and intensity of the tics and may produce side effects (55, 57). Such side effects include sedation, apathy, extrapyramidal effects, weight gain and metabolic abnormalities (for example, Risperidone conveys risks of metabolic syndrome, and weight gain is prevalent) (4, 15, 55, 57)). Because of these risks, it is common practice to postpone tic pharmacological treatment (16, 55). Interventional therapies such as botulinum toxin or deep brain stimulation are only considered in very refractory and severe cases (1, 13, 17, 55).

Even though psychoeducation, psychological and pharmacological treatments are evidence-based and recommended interventions for the treatment of tic disorders, there is still an additional need to develop more global tic-related interventions (55, 60). Consideration must be given to a sensory-based treatment approach that compliments existing treatments for children with tics and comorbidities. Due to the 'sensory phenomena' often described by people with tics, including premonitory urge, 'just-right' perceptions or somatic hypersensitivity associated with tic disorders, novel approaches, such as those that target sensory symptoms, may be useful in children with tic disorders (1). Using a sensory-motor-

based treatment approach has not previously been trialled with children with tic disorders to reduce tic frequency and intensity.

2.2 Sensory Dysregulation and Neurodevelopmental Disorders

2.2.1 Terminology and Broad Overview of Sensory Dysregulation

Dr A. Jean Ayres, an occupational therapist, first defined the term ‘‘sensory integration dysfunction’’ in 1963 (5). Dr Ayres' work explored the interplay between sensory processing and the child’s behavioural response, theorising that impaired sensory processing might result in various functional impairments (5). Occupational therapists have adopted many terms in the literature to describe or explain observable behaviour responses to sensory stimuli (5). For instance, occupational therapists may describe a child as having a ‘‘low threshold to sound’’ after observing a child covering their ears due to loud noise or not using public toilets due to the sound of flushing toilets and hand dryers (5). But neurophysiologists may discuss the term ‘‘threshold’’ concerning a physiological process that denotes the level at which synaptic activity occurs within the central nervous system in response to a stimulus (5).

There are many terms used to describe the sensory features of neurodevelopmental disorders, including ‘‘sensory dysregulation’’, ‘’sensory regulation’’, ‘‘sensory phenomena’’, ‘‘sensory modulation disorder’’, ‘‘sensory processing’’, ‘‘sensory reactivity’’ and ‘‘atypical sensory reactivity’’ (2, 5, 7-9). Due to the many terms used, clinicians lack clarity and consistency within the occupational therapy profession and across disciplines and professionals within the health industry.

For the purposes of this literature review, the popularised term ‘sensory dysregulation’ will be used (5, 7, 9, 10). The developmental psychology literature extensively uses the concepts of emotional (61-63), behaviour (62), and attention regulation (64) and therefore, including the

term sensory regulation is fitting and parallels this concept (9). The National Center of Clinical Infant Programs (NCCIP) defines sensory and motor processing under the conditions termed ‘‘regulatory disorders’’ (65). According to the Diagnostic Classification of Mental Health and Developmental Disorders of Infancy and Early Childhood, regulatory disorders are an inability to regulate behaviours and responses in the domains of physiology, sensation, attention, motor and effective processing (66). Difficulties with children being able to be organised, calm and alert (emotionally regulated) can result from issues in modulating and integrating physiological, sensory, motor and attentional processes (67). Therefore ‘‘sensory dysregulation’’ can lead to emotional dysregulation and is a fitting term for the difficulties children experience with sensory regulation.

2.2.2 Prevalence of Sensory Dysregulation Symptoms in Neurodevelopmental Disorders

Sensory dysregulation is common in people with neurodevelopmental disorders (68-72) and is associated with functional impairment and decreased participation in activities of daily life (73-76). It is believed that less than 13.7% of children commencing school will experience sensory dysregulation problems (77). Sensory dysregulation is a recognised diagnostic feature of autism spectrum disorder (78). However, children with other NDDs also experience sensory dysregulation. For instance, increased sensory dysregulation has also been reported in people with OCD (79-82) and ADHD (10, 18, 68, 72, 83-92). In addition to the premonitory urge mentioned earlier, children or adults with tic disorders and other comorbid NDDs experience sensory dysregulation (25, 93-96).

The literature was reviewed in relation to studies investigating sensory dysregulation symptoms in children (birth to eighteen years of age) with neurodevelopmental disorders using proxy-reported sensory assessment measures (Chapter 2, Table 2A-E). Sensory dysregulation

is more prevalent in children with neurodevelopmental disorders than in typically developing children (39,49,66,67,73,79,81,99). Sensory dysregulation has been extensively researched in children with ASD (8, 22, 24, 73-76, 78, 97-131). Children with ADHD were identified as experiencing increased sensory dysregulation symptoms (85, 86, 91) and 81% more likely in the presence of fetal alcohol syndrome (68). Abnormal sensory dysregulation was also found to have a significant relationship with overall adaptive behaviour (127). As was the case with children with ADHD, children with OCD were also reported to experience increased sensory dysregulation symptoms (79, 80)

Yet, at the time of commencing this PhD in 2017, there was only a single study into sensory dysregulation in children with tic disorders (25). Since this study by Jewers et al. (2013) (25) which investigated sensory dysregulation in children with TS and ADHD compared with children with only TS, there has been growing interest in the area of sensory dysregulation in both children (7, 83, 132, 133) and adults (95, 134, 135) with tic disorders. Furthermore, there is a growing interest globally in research into sensory dysregulation in children with various neurodevelopmental disorders, with research occurring in Australia (22, 99, 100), Brazil (72), Canada (25, 68, 90), Iran (88), Israel (7, 87, 92), Taiwan (18), Turkey (81), the United Kingdom (77, 132), and the United States of America (77, 85, 86, 91, 119, 123, 136). With growing interest in the area of sensory dysregulation in children with tic disorders, a recent case series study in 2018 was the first article to report on misophonic experiences and associated clinical characteristics in young people with tic disorders (133). Misophonia is the triggering of inappropriate or extreme emotional or psychological responses as a result of certain sounds. This study by Robinson et al. (2018) highlights misophonia could be an underestimated phenomenon for abrupt emotional dysregulation in children with tic disorders (133).

Table 2: Provides a list of the details and finding from studies investigating sensory dysregulation in various neurodevelopmental conditions utilising proxy-reported sensory-based measures

Table 2A

Sensory Dysregulation and Neurodevelopmental disorder studies in paediatric populations							
Reference	**Purpose of Study**	**Country**	**Healthcare setting/Centres Involved**	**Study Population** Age: Range [R], and/or Mean [M], Standard deviation [SD] & Gender details	**Control Group** Age: Range [R], and/or Mean [M], Standard deviation [SD] & Gender details	**Assessment Measures Used**	**Findings**
Ahn (2004) (77)	To systematically examine estimated rates of sensory processing disorders using survey data.	USA	Kindergartens from one suburban USA public school district	**n=703** Some children may have had some level of disability. 1% of the sample had special needs	Nil	**Short Sensory Profile (SSP)**	Ninety-six children (13.7 %) met criteria for sensory processing disorders based on parental perceptions. These percentages are consistent with hypothesised estimates published in the literature.
Rogers, Hepburn & Walker (2002) (127)	This study sought to examine the pervasiveness of behaviours thought to reflect sensory abnormalities that parents report in toddlers with autism	USA	Various health & early education agencies	**n=78** **ASD: n=26.** Age range: 26-41 months, M=33.67 months. SD=3.6 months **Fragile X syndrome: n=20** Age range: 21-50 months, M=36.11 months. SD=8.1 months & **Developmental Delay**: n=32 (Down syndrome: n=15; premature birth: n=4 & significant medical history n=4. Age range: 24-47 months, M=33.23 months SD=6.7 months Gender not reported	**n=24** TD* children. Age range 12- 35 months. Mean age 19 months. SD=4.8 months Gender not reported	**SSP** ADI-R, ADOS, Vineland Scales of adaptive behaviour Interview.	Children with Fragile X syndrome & ASD had significantly more sensory symptoms overall than the two comparison groups. Children with ASD and Fragile X syndrome were more impaired than developmentally delayed & TD in tactile sensitivity & auditory filtering. Children with ASD had more difficulty with taste & smell. Abnormal sensory reactivity had a significant relationship with overall adaptive behaviour.

Table 2B

Sensory Dysregulation and Attention Deficit Hyperactivity Disorder/ Attention Deficit Disorder (ADHD/ ADD) studies in paediatric populations							
Reference	**Purpose of Study**	**Country**	**Healthcare setting/Centres Involved**	**Study Population** Age: Range [R], and/or Mean [M], Standard deviation [SD] & Gender details	**Control Group** Age: Range [R], and/or Mean [M], Standard deviation [SD] & Gender details	**Assessment Measures Used**	**Findings**
Abele-Webster (2012) (68)	To understand sensory processing in these children, which may assist with early identification & intervention.	Canada	Canadian rehabilitation hospital	**n=26** Age range: 5-10 Children with FASD# & ADHD. Mean age & gender not provided.	Nil	**SSP,** CPRS-R: L	Sensory processing problems were found in 81% of the children, similar to other studies of children with fetal alcohol spectrum disorder.
DeSerisy (2019) (85)	It was hypothesised that sensory sensitivity would moderate the relationship between hyperactive-impulsive symptoms of ADHD & emotional lability in these youth.	USA	School referrals, mental health clinics, flyers & newspapers & Internet advertisements.	**n=24** Low & Typical sensory sensitivity M = 7.96 yrs. SD = 1.35 yrs. Male: n= 18, Female: n=6. Hyperactive/impulsive symptoms: n=3.75	n=47 High sensory sensitivity M = 7.67 yrs. SD=1.02 yrs. Male: n=37, Female: n=10 Hyperactive/impulsive symptoms: n=4.91	**SSP** Emotional regulation checklist & KSADSPL	Results indicate that heightened sensory sensitivity increases hyperactive/impulsive symptoms of ADHD & emotional lability in children with three or more clinically impairing ADHD symptoms. This study adds to the growing evidence that children with sub-threshold ADHD experience significant functional impairment & high rates of sensory sensitivity.
Dunn (2002) (86)	To compare the sensory responses of children with attention deficit hyperactivity disorder (ADHD) & children without disabilities on the Sensory Profile.	USA	Community ADHD clinic.	**n=70** Age range: 3-15 yrs. Mean age not provided Male: n=61, Female: n=9 n=52 taking ADHD medication	**n=70** Age and gender matched without disabilities from the previous study. Male: n=61, Female: n=9	**SP** Demographic sheet	Children with ADHD differed significantly from children without disabilities in their sensory responsiveness based on Sensory Profile results.
Pfeiffer (2015) (91)	The study aimed to investigate whether children with ADHD are at greater risk than children without ADHD for sensory processing problems.	USA	Recruited from a paediatrician's office, an ADHD information fair; & a Web site specific to childhood ADHD.	**n=20** Children with ADHD Age range: 5-10 yrs. M = 9.1yrs., SD =1.3 yrs. Male: n=15, Female: n=5	**n=27** TD Children. Age range:5-10 yrs. M = 8.3 yrs. SD= 1.6 yrs. Male: n=13 Female: n=14	**SPM-Home Form** Conners-P	Children with ADHD exhibited more sensory processing problems on all scales of the Sensory Processing Measure with small to medium effect sizes observed ($\eta 2$ = .27 to .61).

Table 2C

Sensory Dysregulation and Autism Spectrum Disorder (ASD) studies in the paediatric population							
Reference	**Purpose of Study**	**Country**	**Healthcare setting/Centres Involved**	**Study Population** Age: Range [R], and/or Mean [M], Standard deviation [SD] & Gender details	**Control Group** Age: Range [R], and/or Mean [M], Standard deviation [SD] & Gender details	**Assessment Measures Used**	**Findings**
Asburner (2008) (22)	Explored the associations between sensory processing & classroom emotional, behavioural and educational outcomes of children with ASD.	Australia	Recruited from 12 Queensland schools.	**n=28** Children with ASD Males: n=24 Female: n=4	**n=51** TD children Males: n=43 Females: n=8	**SSP** GARS GADS K-BIT CTRS-R: L ASEBA: TRF	Significant group differences were found on all SSP factor scores & total scores (p<.001) with the exception of Movement Sensitivity (t=-2.46, p=.016, df=57.98). Seventy-nine percent of children with ASD had SSP total scores in the definite difference (DD), compared with 2% DD in the TD group.
Brockevelt (2013) (98)	To further investigate these differences among children ages 3 to 9 years as measured by the SP.	USA	University of Dakota diagnostic clinic	**n=21** Children with ASD Age range 3-9 yrs.	**n=21** Age & gender-matched TD Age range 3-9 yrs.	**SP**	Significant differences were found across all four SP quadrants in ASD group (Registration, Seeking, Sensitivity, & Avoiding), as well as eight of the nine SP factor scores.
Kientz (1997) (116)	To determine whether the Sensory Profile discriminates between children with and without autism and which items on the profile best discriminate between these groups.	USA	Northwest Missouri Autism Consortium	**n=32** children with autism Age range: 3-13 yrs. Male: n=26, Female: n=6	**n=64** Without autism Age ranged: 3-10 yrs.	**SP** CARS	Eight-four of 99 items (85%) on the Sensory Profile differentiated the sensory processing skills of subjects with autism from those without autism.
Tomcheck (2007) (8)	To investigate differences in sensory processing among age-matched children between ages 3 and 6 years with ASD & TD children.	USA	Tertiary Diagnostic centre	**n=281** children with ASD Male: n=235, Female: n=46	**n=281** age-matched TD peers Male: 235, Female: n=43	**SSP**	Ninety-five percent of children with ASD demonstrated sensory processing dysfunction on the SSP Total Score, with the greatest differences reported on the Under responsive/ Seeks Sensation, Auditory Filtering, & Tactile Sensitivity sections.
				Mean age of total sample: 51.58 months, SD=10.3 months			

Table 2D

Sensory Dysregulation and Obsessive-compulsive disorder (OCD) studies in the paediatric population							
Reference	**Purpose of Study**	**Country**	**Healthcare setting/Centres Involved**	**Study Population** Age: Range [R], and/or Mean [M], Standard deviation [SD] & Gender details	**Control Group** Age: Range [R], and/or Mean [M], Standard deviation [SD] & Gender details	**Assessment Measures Used**	**Findings**
Lewin (2015) (82)	To examine sensory over-responsivity (SOR) in a clinical sample of youth with OCD.	USA	Specialty outpatient treatment program	**n=80** Youth with OCD Age range:3-18 yrs. M:8.91, SD: 3.91 yrs. Male: n=50, Female: n=30 Comorbid diagnoses: anxiety disorders (56.3 %) and/or ADHD (31.3 %). n=31 prescribed psychotropic medications.	Nil	**SSP** CGI-S, CYBOCS, ADIS-IV-C/P, CSDS	Sensory over-responsivity (SOR) is common among youth with OCD; 32.5 % experienced tactile hypersensitivity, 20.3 % experienced visual/ auditory hypersensitivity, & 20.5 % experienced gustatory/ olfactory hypersensitivity. SOR was more common in younger children.
Dar (2011) (80)	To investigate the hypothesis that strong reactions to everyday sensory events are related to ritualism in children.	Israel	Recruited from acquaintances of the experimenter	**n=61** Children with OCD Age range 4-6.4 years Male: n=39, Female: n=22	Nil	**SP** CRI, SCARED	Strong reactions to everyday sensory events were highly correlated with childhood ritualism, even after controlling for anxiety.

Table 2E

Sensory and Tic Disorder studies in paediatric populations							
Reference	**Purpose of Study**	**Country**	**Healthcare setting/Centres Involved**	**Study Population** Age: Range [R], and/or Mean [M], Standard deviation [SD] & Gender details	**Control Group** Age: Range [R], and/or Mean [M], Standard deviation [SD] & Gender details	**Assessment Measures Used**	**Findings**
Jewers (2013) (25)	To determine whether sensory processing differences existed in children aged 5 & 10 years with TS.	Canada	Tertiary care hospital with a specialised TD program	**n=28** Children with tic disorders Age range for entire sample n=75: 5.6 to 10.9 yrs. M=9.05, SD=1.31 yrs. Male: n=71, Female: n=4.	**n=47** Children with tic disorders & ADHD	**SSP** Demographic survey	Children with tic disorders and ADHD experienced more significant sensory dysregulation difficulties than children with only tic disorders.
Weisman (2017) (7)	The aim of the study was to investigate sensory dysregulation as a feature of TS & its related conditions.	Israel	Tertiary Tourette syndrome Clinic	**n=92** Children with TS/ chronic tic disorder. Age range 7-14 yrs. Male: n=78, Female: n=14	Nil	**SSP FPT, Von Frey Filament Test**, YGTSS, PUTS, CY-BOCS, CDI, Conners-P, SCARED & QoL questionnaire.	Sensory dysregulation was significantly more common & severe when there were comorbidities. The presence of SMD was associated with more severe impairments in quality of life & less participation in daily activities.

ADHD = Attention Deficit Hyperactivity Disorder, ASD= Autism spectrum disorder, OCD= Obsessive-compulsive disorder, TD= Typically developing, FASD = Fetal Alcohol syndrome disorder

List of all the abbreviations of sensory assessment measures listed in the tables: Sensory Processing Measure-Home Form (SPM), Short Sensory Profile (SSP), Sensory Profile (SP).

List of all the abbreviations of all other assessment measures listed in the tables in alphabetical order: Achenback System of Empirically Based Assessment: Teacher Report Form (ASEBRA:TRF) [Achenbach and Rescorla, 2010], Anxiety Disorders Interview Schedule for DSM-IV: Child and Parent Versions (ADIS-IV-C/P) [Scahill et al 1997], Childhood Autism Rating Scale (CARS) [Schopler, Reichler and Renner, 1988], Clinical Global Impression—Severity (CGI-S), Conners' parent questionnaire (Conners-P), Conners' Parent Rating Scales–Revised, Long Version (CPRS-R:L) [Conners, 1997], Childhood Routine Inventory (CRI)[Evans et al.'s 1997], Child Sheehan Disability Scale (CSDS) [Whiteside 2009], Children's yale brown obsessive compulsive scale (CYBOCS), Gilliam Autism Rating Scale (GARS) [Gilliam 1995], Fabric Prickliness Test (FPT), Gilliam Asperger's Disorder Scale (GADS) [Gilliam 20021], Kaufman Brief Intelligence Test (K-BIT) [Kaufman and Kaufman 1990], & Kiddie Schedule for Affective Disorders and Schizophrenia (KSADSPL) [Kaufman, Birmaher, Brent, and Rao, 1997], Premonitory urge for tics scale (PUTS), Quality of life (QoL) child/parent questionnaire, Screen for child anxiety-related emotional disorders (SCARED).

From electrophysiological magnetoencephalography (137), functional imaging (138) and volumetric imaging (139) studies, it has been shown that there is substantial convergent evidence of sensorimotor abnormalities in people with tic disorders and TS (140, 141). Deficits in sensorimotor gating have been identified in individuals with TS, resulting in problems filtering irrelevant sensory stimuli (140, 142). For these and the reasons mentioned above, tics are considered a 'sensorimotor' phenomenon rather than a pure movement disorder (7, 141).

The severity of sensory dysregulation symptoms has been consistently demonstrated to increase with the presence of comorbidities in children with neurodevelopmental disorders (7, 25, 82). This is key to understanding sensory dysregulation symptoms in children with tic disorders, with 80-90% of these children experiencing other comorbidities.

2.2.3 Functional Impairment of Sensory Dysregulation in Children

Sensory dysregulation can result in functional impairment and impact children's participation and enjoyment of tasks due to their atypical reactions to sensory stimuli (143). Difficulties associated with sensory dysregulation across various neurodevelopmental disorders have been categorised into five functional impairment areas: (i) decreased social skills and involvement in occupational performance areas; (ii) reduced frequency, duration, or complexity of adaptive responses; (iii) impaired self-confidence or self-esteem; (iv) poor family and daily life skills; and (v) impaired gross-motor, fine-motor and sensory-motor skill development (144). Sensory dysregulation is also associated with decreased school participation and increased parental stress (70, 73, 143, 145).

We have anecdotally observed that patients with tics frequently report sensory dysregulation symptoms that lead to functional impairment in their ability to engage in daily activities. Common symptoms include tactile sensitivities such as intolerance of clothing tags and fabrics,

intolerance to auditory stimuli such as noise in busy shopping centres, chewing or other human-made noises, and temperature sensitivity. These sensory dysregulation symptoms can result in behavioural changes such as emotional responses to stimuli. They are also associated with functional impairments such as the inability to wear seat belts, wear school clothes or attend social activities. These problems result in stress for the child and family.

2.2.4 Measures Used to Assess Sensory Dysregulation in Children

Assessment and management of sensory dysregulation is an accepted part of comprehensive care for children with NDDs (146). For clinical and research purposes, there are two commonly used parent-reported questionnaires developed for neurodevelopmental disorders to assess sensory dysregulation in children. These are the Sensory Processing Measure (SPM) (147) and the Child Sensory Profile-2 (CSP2) (148). There is an abbreviated version of the CSP2, the Short Sensory Profile 2 (SSP2) (8), which, although having the same questions, has a reduced number of questions and is frequently used for research. These questionnaires use a Likert scale with high scores denoting more significant impairment.

The SPM (75 questions) provides information on sensory dysregulation by giving a total score and sub-scores for vision, hearing, touch, taste and smell, body awareness and balance and motion for children between the ages of 5 and 12 years of age (26). The questionnaire also provides a score for social participation and planning, and ideas (26). Even though this tool is believed to be a valid and reliable measurement tool for children with neurodevelopmental disorders (26), it has not previously been used in studies in a tic population.

The Child Sensory Profile-2 (CSP2) was developed in 2014, following this measure superseding the Child Sensory Profile (CSP) published in 1999. The CSP2 evaluates sensory patterns or dysregulation in the context of daily functional tasks for children between the ages

of 3 and 14 years (100, 148). This proxy-reported measure has 86 items and takes approximately 15 to 20 minutes to complete by the parent or carer of the child (148). In terms of scoring, the Sensory Profile-2 provides only sub-scores of all sensory areas and not a total score (148). On the other hand, the Short Sensory Profile-2 (SSP2), which too replaces the Short Sensory Profile (SSP) as of 2014, has 34 items identical to the CSP2 and takes approximately 5 to 10 minutes to complete but provides two total scores, an overall Sensory Process score and a Behavioural Responses score (8, 148). There are no individual sensory domain scores for each of the senses on the SSP2 as there are for the CSP2. Therefore, using these measures simultaneously allows for both subscore/domain scores for each sensory area obtained from the CSP2 and overall total scores gained from the SSP2 (148).

In all the studies identified and those highlighted in Table 2 above, the Sensory Profile and an abbreviated version of the same measure, the Short Sensory Profile (149), are most commonly used as the assessment measure to evaluate sensory dysregulation in children. Only one study (91) uses the Sensory Processing Measure-Home Form (26).

2.2.5 Therapeutic Approaches for Sensory Dysregulation in Children

Therapeutic approaches are commonly used to address sensory dysregulation in children with NDDs, with most of these strategies having been developed for children with ASD (8, 150-153). Sensory dysregulation treatment approaches include but are not limited to the Wilbarger Protocol (20), the Therapeutic Listening Program (21), the Astronaut Training Program (19) and the Alert Program®. The Wilbarger Program uses a specific therapressure brush to apply mid-range pressure to the person's back, arms and legs, and in addition, joint compressions are provided (20). The Wilbarger Protocol is the most popular used program to treat sensory over-responsivity in children between 2 and 12 years (20). The Therapeutic Listening Program is an auditory intervention approach to reduce auditory sensitivity through the child listening to

electronically altered music using specific headphones twice daily as a home program (21). The Astronaut Training Protocol uses rotation, inversion and linear movement to stimulate the vestibular, visual and auditory systems (19). The evidence indicates that the Astronaut Training Protocol may assist with vestibular function and postural control in children (19).

The Alert Program® is a treatment approach that uses sensory-motor strategies to assist children and adults with self-regulation and attention skills (29). Self-regulation is believed to be comprised of the management of physiological arousal, emotions, attention and behaviour, and is required in order to perform optimally in daily activities (28). As children with tic disorders were identified as experiencing sensory dysregulation symptoms (25) and emotional dysregulation exacerbates tics (12), trialling the Alert Program® of all the sensory programs to manage tic seemed to be the best fit for our planned intervention.

The Alert Program's® primary aim is to teach children awareness of their emotional dysregulation state (or level of arousal as referred to by the program) and then to monitor, maintain and change from emotional dysregulation to emotional regulation, using strategies taught by the trained therapist (29). The program consists of lessons and activities incorporating both sensory-motor strategies and cognitive approaches (29). The program is tailored to the child's specific sensory preferences and is intended to be utilised in conjunction with other therapies, medical practices, and treatments (29). The program is broken into strategies that can assist with changing how alert the child feels across the different senses, i.e., i) put something in your mouth (oral input), ii) move (vestibular input), iii) touch (tactile input), iv) look (visual input) and vi) listen (auditory input)) (29).

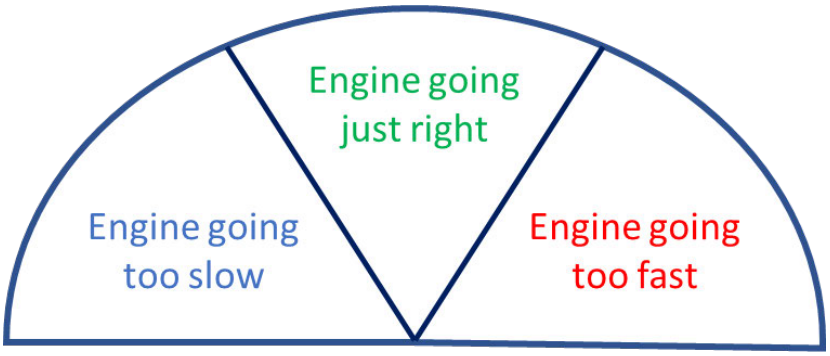

Figure 2: Diagram depicting the speedometer analogy of the Alert Program® to explain the different levels of emotional dysregulation (engine going too fast or too slow) or emotional regulation (engine going just right) (29).

Similar to the Alert Program®, The Zones of Regulation® is another program tailored to the individual student and aims to teach self-regulation skills (154). The Zones of Regulation® uses a systematic cognitive-behavioural approach to teach self-regulation skills through eighteen lessons (154). The program aims to i) expand students' vocabulary of emotional terms, ii) learn to read facial expressions, iii) gain an understanding of how others see and react to their behaviour, iv) identify emotional triggers and v) increase their problem-solving skills (154). The program uses (a) calming techniques/mindfulness strategies, (b) cognitive or thinking strategies, and (c) sensory supports (154). Neither the Alert Program® nor the Zones of Regulation® program have been trialled in the management of tic disorders with children with tic disorders.

As emotional dysregulation (i.e., stress, anxiety, excitement and fatigue) exacerbates tics (12), a treatment approach that provides emotional regulation strategies may be effective in tic reduction. It is also understood that children with tic disorders and comorbidities experience sensory dysregulation (7, 25). As sensory-motor strategies are fundamental to the framework of The Alert Program® to gain emotional regulation, it was believed to be a better fit for this

population. Therefore, implementing the Alert Program®, which uses sensory-motor strategies to manage self-regulation, may be a good fit in terms of a treatment approach for this population.

CHAPTER THREE

Sensory Dysregulation in Childhood Tic Disorders is Associated with Executive Dysfunction and comorbidities

Although we planned to pilot a sensory-based treatment approach, the Alert Program® with children with tic disorders to reduce tics, it was imperative that we first understood the prevalence of the sensory dysregulation symptoms in our population before commencing treatment. This chapter provides information on the first study we conducted to understand the prevalence of sensory dysregulation symptoms in Australian children with a tic disorder attending a tertiary-level paediatric hospital service. As this study was published, this chapter is presented in the published format of the paper. A preface to the chapter, as well as an authorship statement, has been provided. Thereafter the sections: abstract, background, methods, results, and discussion will follow in the format of the journal *Movement Disorders*. A list of references will be found at the end of the chapter. Supplementary material referred to in the manuscript is provided in the appendix section of the thesis.

Preface

At the time of commencing our prevalence study, there had only been a single study investigating sensory dysregulation in children with tic disorders (25). The Canadian study compared the prevalence of sensory dysregulation symptoms in children with tic disorders in the presence and absence of ADHD (25). Although 54-60% of children with tic disorders have a co-existing diagnosis of ADHD, children with tic disorders are afflicted by other neurodevelopmental and neuropsychiatric conditions such as OCD (30-40%), obsessive-compulsive behaviours (OCB) (60-90%), ADHD (54-60%), ASD (5-15%) and depressive disorders (13-76%) (4, 13, 40, 41). Therefore there was a gap in the literature to provide an understanding of the prevalence of sensory dysregulation symptoms in children with tic disorders and various neurodevelopmental disorders and not only in children with tic disorders in the presence or absence of ADHD.

Through our first study and this chapter, we aimed to investigate sensory dysregulation in children with tic disorders using commonly used proxy-report sensory-based measures, the Child Sensory Profile-2 (CSP2) and the Sensory Processing Measure (SPM), to determine the prevalence of sensory dysregulation in children with tic disorders and comorbidities. The CSP-2 measure has 86 items and takes approximately 15 to 20 minutes to complete by the parent or carer of the child (148). In terms of scoring, the Sensory Profile-2 provides only sub-scores of all sensory areas and not a total score (148). On the other hand, the Short Sensory Profile-2 (SSP2), which also replaces the Short Sensory Profile (SSP) as of 2014, has 34 items identical to the CSP2 and takes approximately 5 to 10 minutes to complete but provides two total scores, an overall Sensory Process score and a Behavioural Responses score (8, 148). There are no individual sensory domain scores for each of the senses on the SSP2 as there are for the CSP2. Therefore, using these measures simultaneously allows for both subscore/domain scores for

each sensory area obtained from the CSP2 and overall total scores gained from the SSP2 (148). The SPM was included in this study as an additional proxy-report sensory-based measure as there had been no published study using this assessment with a paediatric tic population, and the researcher was interested to know if this was an appropriate tool to consider. As there was no significant difference in the results between the two sensory measures (CSP2 and the SPM) with children with tic disorders, only the results from the CSP2 and SSP2 were reported in the published paper. The comorbidities of the study participants with tic disorders involved in this research study included anxiety disorders, ADHD, ASD, depressive disorders, intellectual disability and OCD. Therefore, this study was broader and more comprehensive than the study conducted by Jewers et al. (2013) and examined the 'real life' complex comorbidity that is so common in tic disorders (25).

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Soler. N, Hardwick. C, Perkes. I., Mohammad. S., Dossetor. D., Nunn, K., Bray. P, Dale RC. Prevalence of sensory symptoms and emotional regulation in Australian children with tic disorders. ***European Society for the study of Tourette's syndrome Conference (ESSTS). Copenhagen, Denmark. June 2019***.

Soler. N, Hardwick. C, Perkes. I., Mohammad. S., Dossetor. D., Nunn, K., Bray. P, Dale R. Sensory dysregulation in tic disorders is associated with executive dysfunction and comorbidities. ***The University of Sydney Children's Hospital Westmead Clinical School Discipline of Child and Adolescent Health, Higher Degree Research Conference, Parramatta, the 16th of August 2019.***

National Conferences

Soler. N, Hardwick. C, Perkes. I., Mohammad. S., Dossetor. D., Nunn, K., Bray. P, Dale R. Prevalence of sensory and emotional regulation symptoms in Australian children with tic disorders. ***The University of Sydney Children's Hospital Westmead Clinical School Discipline of Child and Adolescent Health, Higher Degree Research Conference, Parramatta. August 2018***.

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Authorship Statement

The co-authors of the paper *''Sensory dysregulation in tic disorders is associated with executive dysfunction and comorbidities*''confirm that ***Nicolette Soler*** has made the following contributions:

- Conception and design of the research
- Collection and extraction of all data
- Analysis and interpretation of the findings
- Drafting and revising the manuscript and critical appraisal of content

As the primary supervisor for the candidature upon which this thesis is based, I can confirm that the above authorship attribution statement is correct.

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Date: 28-05-2023

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Date: 28-05-2023

RESEARCH ARTICLE

Sensory Dysregulation in Tic Disorders Is Associated With Executive Dysfunction and Comorbidities

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ABSTRACT: **Background**: Tics are conceptualized as a sensorimotor phenomenon with a premonitory urge typically described by patients. As observed in other neurodevelopmental disorders, we have observed sensory dysregulation symptoms, such as tactile hypersensitivity to clothing, in children with tic disorders; however, formal clinical research in this area is limited.

Objective: To define the presence of sensory dysregulation symptoms in tic disorders, and their clinical associations.

Methods: Prevalence of sensory dysregulation in 102 children with tic disorders was compared to 61 age- and sex-matched healthy controls. Sensory dysregulation, executive function, and quality of life data were obtained through the Short Sensory Profile-2, Sensory Profile-2, Sensory Processing Measure, Behaviour Rating Inventory of Executive Function-2, and Strength and Difficulties Questionnaire and Pediatric Quality of Life Inventory. Tics were assessed with the Yale Global Tic Severity Scale.

Results: Children with tics, in the presence of comorbidity, had elevated sensory dysregulation compared to healthy controls ($P < 0.001$). There was a positive correlation between sensory dysregulation and global executive difficulties in children with tics and comorbidity (n = 87; rho = 0.716; $P < 0.001$) and a negative correlation of sensory dysregulation with quality of life (n = 87; rho = –0.595; $P < 0.001$). In children with tics, there was an association between sensory dysregulation and number of comorbidities ($P < 0.001$).

Conclusion: In the presence of comorbidity, children with tic disorders have broad sensory dysregulation symptoms beyond the premonitory urge. There was a statistically significant association between sensory dysregulation and executive function difficulties and the presence of neurodevelopmental and psychiatric comorbidity. Sensory dysregulation can be considered neurodevelopmental symptoms, providing insight into the neurobiology of tics and opportunities for therapeutic intervention. © 2019 International Parkinson and Movement Disorder Society

Key Words: child; executive dysfunction; neurodevelopmental disorders; sensory dysregulation; tic disorders

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Tics are repetitive, stereotypical, rapid, nonrhythmic movements or vocalizations. Tic disorders are the most common childhood movement disorder and regularly coexist with other neurodevelopmental problems.[1,2]

"Sensory phenomena" often described by people with tics include premonitory urge, "just-right" perceptions, or somatic hypersensitivity.[4] Premonitory urge is a discomfort or tension that people with tics experience immediately before a tic and it may play a role in tic generation.[3,4] In addition to this sensory experience, broader sensory dysregulation has been described in people with tics.[5-10]

For a person to engage in daily tasks effectively while adapting to challenges, there needs to be the "'capacity to regulate and organize the degree, intensity and nature of the responses to sensory input in a graded and adaptive manner."[11] Impaired ability to manage sensory input results in "behavioral responses disproportional to the sensory input experienced."[12]

Terms to describe the sensory features of neurodevelopmental disorders include sensory dysregulation, sensory phenomena, and atypical sensory reactivity.[7,11,13,14] Here, we have used the popularized term "sensory dysregulation."[7,15,16]

Sensory dysregulation can impact on children's participation and enjoyment of tasks because of their atypical reactions to sensory stimuli.[17] Difficulties associated with sensory dysregulation were categorized into five functional impairment areas: (1) decreased social skills and involvement in occupational performance areas; (2) reduced frequency, duration, or complexity of adaptive responses; (3) impaired self-confidence or self-esteem; (4) poor family and daily life skills; and (5) impaired gross-motor, fine-motor, and sensory-motor skill development.[18]

We have observed that patients with tics frequently report functionally impairing sensory dysregulation. Common symptoms include tactile sensitivities, such as intolerance of clothing tags and fabrics, intolerance to auditory stimuli such as noise in busy shopping centres, chewing or other human-made noises, and sensitivity to temperature. These sensory dysregulation symptoms can result in behavioural change, such as emotional responses to stimuli. They are also associated with functional impairments, such as inability to wear seat belts, wear school clothes, or attend social activities. These problems result in stress to the child and family.

There is substantial convergent evidence of sensorimotor abnormalities in people with tic disorders and Tourette's syndrome (TS) from electrophysiological studies,[13,19] magnetoencephalography,[20] functional imaging,[21] and volumetric imaging.[22] Individuals with TS have been described as experiencing deficits in sensorimotor gating, which result in problems filtering irrelevant sensory stimuli.[5,19] For these and the above-mentioned reasons, tics are considered a "sensorimotor" phenomenon, rather than a pure movement disorder.[7,13]

Other neurodevelopmental disorders typically co-occur with tic disorders, including attention-deficit hyperactivity disorder (ADHD), childhood-onset obsessive-compulsive disorder (OCD),[23] and autism spectrum disorder (ASD). Sensory dysregulation are common in people with neurodevelopmental disorders,[24-29] particularly ASD, and are associated with impaired function.[30-34] Sensory dysregulation is associated with decreased school participation and engagement in daily tasks and increased parental stress levels.[17,26,30,35] Sensorimotor-based therapy is a method to manage children with autism with sensory dysregulation.[14,36-38]

In the current study, we aimed to investigate sensory dysregulation in children with tic disorders, using validated tools for sensory regulation in children with neurodevelopmental disorders, the Sensory Profile-2 (SP2)[39] and the Sensory Processing Measure (SPM).[40] We hypothesized that sensory dysregulation would be present in children with tic disorders and have a dose-response relationship relative to neurodevelopmental and psychiatric comorbidities. Furthermore, we hypothesized that sensory dysregulation would correlate with other executive symptoms, particularly emotional dysregulation.

Participants and Methods

Participants

Participants were recruited in this cross-sectional case control study at the Children's Hospital at Westmead in Sydney, Australia between January 2017 and April 2018, after ethics approval (LNR/17/SCHN/8). Inclusion criteria for all groups required the parents to speak conversational English and for their child to be aged between 5 years 0 months and 12 years 11 months. This age range is the validated age range of the SPM.[40] Participants with existing comorbidities were included in the study.

The participants in the study were children with diagnosed tic disorders recruited through the outpatient tic clinic at the Children's Hospital at Westmead. This is a multidisciplinary clinic at the largest referral service for children with tic disorders in Western Sydney. Clinic patients were referred by general practitioners and pediatricians. One hundred twenty-three consecutive tic clinic patients were invited to participate, of which 102 participants completed the study (71 males, 31 females; median age range was 9 years and 5 months, range 5 years to 12 years 11 months). The tic disorder cohort included children with TS (n = 82; 80.4%), provisional tic disorder (n = 13; 12.7%), chronic vocal tic disorder (n = 1; 1%), and chronic motor tic disorder (n = 6; 5.9%). Of the 102 participants with tic disorders, 88 were diagnosed with existing Diagnostic and Statistical Manual 5 (DSM-5)[41] criteria comorbidities, which included anxiety (n = 68), OCD (n = 45), ADHD (n = 39), ASD

(n = 27), intellectual disability (n = 20), and depressive disorder (n = 14). We refer to this group as "'children with tic and comorbidities." Only 14 tic participants (n = 5 with provisional tic disorder) were identified with no coexisting comorbidities (referred to as "children with tic only").

To generate normative data, we recruited a control group of typically developing children of hospital staff. As an eligibility-screening method, parents were asked whether their children were diagnosed or suspected of having any disorder of development. Parents who identified neurodevelopmental concerns or diagnosis were not able to participate in the study. Ninety-two control participants were invited to participate, and 61 assessments were completed (44 males, 17 females; median age range was 9 years and 4 months, range 5 years to 12 years, 9 months). The patient and controls were sex and age matched, and there was no significant difference between the two participant groups in ethnicity, handedness, and education.

One tic participant did not complete the SP2 and another did not complete the Behaviour Rating Inventory of Executive Function-2 (BRIEF-2; n = 101 for these questionnaires, although there were 102 tic participants).

Assessment Instruments

There were two data collection stages. Stage 1 was parent-reported outcome measures using five validated questionnaires. Stage 2 used a clinician interview using the Yale Global Tic Severity Scale (YGTSS).

Parents completed five questionnaires (30–45 minutes' duration) before or after their clinician interview. To assess sensory dysregulation, we used two reliable and valid parent-rated questionnaires developed for neurodevelopmental disorders, which assess sensory dysregulation in children, the SPM[40] and the SP2,[39] including the short version of the SP2, the Short Sensory Profile 2 (SSP2).[14] Both questionnaires use a Likert scale, with high scores denoting greater impairment.

The SPM (75 questions) provides information on sensory dysregulation by providing a total score and subscores for vision, hearing, touch, taste and smell, body awareness, and balance and motion for children between the ages of 5 and 12 years. The questionnaire also provides a score for social participation and planning and ideas. Although a valid and reliable tool previously used in neurodevelopmental disorders,[14,15,28] the SPM has not previously been used in studies in a tic population. The researchers were uncertain of the responsiveness of these tools in a pediatric tic population, and thus the Sensory Profile-2[39] was used in conjunction with the SPM.

The SP2 evaluates sensory patterns or dysregulation in the context of daily functional tasks for children between the ages of 3 and 14 years. Given that the SP2 provides only subscores of all sensory areas and not a total score, the SSP2[14] scores were calculated from the parent-completed SP2 questionnaire to derive a total raw sensory score as well.

To determine executive function, parents completed the Behaviour Rating Inventory of Executive Function 2 (BRIEF-2) Parent Form Questionnaire, which has acceptable validity and reliability.[42] The BRIEF-2 comprises 63 questions measuring the following functions: inhibition, self-monitoring, shifting, emotional control, initiation, working memory, plan/organizing, task monitoring, and organisation of materials.

The Strengths and Difficulties Questionnaire (SDQ) was used, given that it is considered effective and valid in screening for general pediatric psychopathology.[43]

To investigate the impact of tic disorders on health-related quality of life, we used the Pediatric Quality of Life Inventory (PedsQL; Copyright © 1998 JW Varni, PhD. All rights reserved; with permission: Mapi Research Trust, Lyon, France). The PedsQL comprises of 23 items and has demonstrated reliability and validity.[44,45]

Finally, a clinician interview using DSM-5 criteria for tic disorders and associated comorbidity was conducted by one of two pediatric neurologists (R.D. or S.M.), expert in pediatric movement disorders. Comorbidities routinely screened for included OCD, ADHD, anxiety, and depressive disorders. Many participants were diagnosed with comorbidities, such as ASD, by other health professionals involved in their care before being seen at this clinic. Where an ASD diagnosis was suspected but not confirmed, a referral to a neuropsychologist was made for further assessment. All comorbidities fulfilled DSM-5 criteria. A pediatric neurologist completed the YGTSS.[46]

Statistical Analysis

Given that data were non-normally distributed, to analyze the difference between sensory dysregulation and executive function difficulties between the participant groups, a Kruskal-Wallis test was used. To determine whether a correlation between sensory dysregulation and executive function difficulties existed, a Spearman correlation test was used. Strength of association between values was interpreted as specified by Cohen[47] with a rho = 0.10 interpreted as a small; rho = 0.30 a moderate, and rho = 0.50 a large correlation. A linear regression test was used to test the association between sensory dysregulation (using the total SSP2 raw scores) and the number of comorbidities as a continuous predictor.

All categorical data were assessed using a nonparametric chi-squared test. $P < 0.05$ was interpreted as significant for all analyses. Analyses were completed using SPSS software (version 25; SPSS, Inc., Chicago, IL).[48]

Results

Sensory Dysregulation

There was strong positive correlation between the SSP2 and SPM sensory assessments (n = 162; rho = 0.842; $P < 0.001$). For clarity, only the results for the SSP2 (and SP2) will be subsequently presented in the text (SPM results in Table 2 only).

When comparing sensory dysregulation (using SSP2 total raw scores), participants with tic disorders (n = 101) had significantly elevated scores compared with healthy controls (n = 61; $P < 0.001$; Fig. 1A). These results were further analyzed. We found that there was a significant difference in sensory dysregulation scores on the SSP2 for participants with tics and comorbidities (n = 87) compared to those children with tics only (n = 14; $P < 0.001$; Fig. 1B). There was no significant difference in sensory dysregulation between children with tics only (n = 14) and healthy controls ($P = 0.349$) (Figure 1B). The presence of comorbidity was associated with elevated sensory dysregulation, as shown for ASD (Fig 1C), ADHD (Fig 1D), OCD (Fig 1E) and any emotional disorder (Fig 1F).

All SP2 subscores (i.e., seeking, avoiding, sensor, bystander, auditory, visual, touch, movement, body position and oral, conduct, social emotional, and attentional) were elevated in the total tic group compared to healthy controls (Table 2).

Executive Function

Tic participants (n = 101) had significantly elevated executive function difficulties (BRIEF-2 Global Executive Composite [GEC]) compared to healthy controls (n = 61; $P < 0.001$; Fig. 2A). There was no significant difference between children with tic only and healthy controls ($P = 0.135$; Fig. 2B).

As with sensory dysregulation, presence of comorbidities was associated with worsening executive function, as shown for ASD (Fig. 2C), ADHD (Fig. 2D), OCD (Fig. 2E), and any emotional disorder (Fig. 2F).

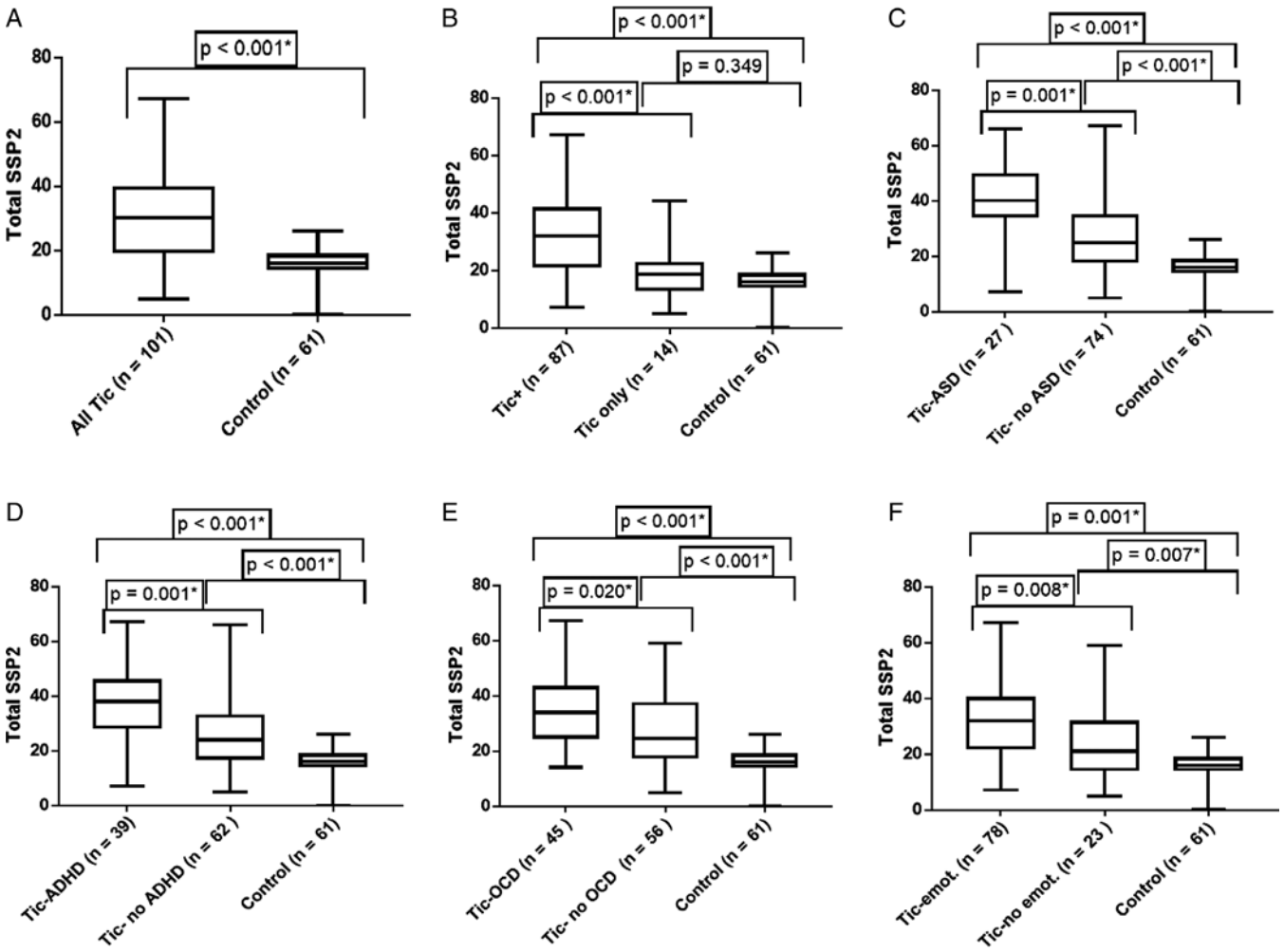

FIG. 1. Sensory dysregulation associated with tic disorders and comorbidity. Note: SSP2 results show elevated sensory dysregulation in tic participants compared to controls (A). Sensory dysregulation were not elevated in the tics-only group (B). The presence of all comorbidities, ASD (C), ADHD (D), OCD (E), and emotional disorder (F) was associated with increased sensory scores.

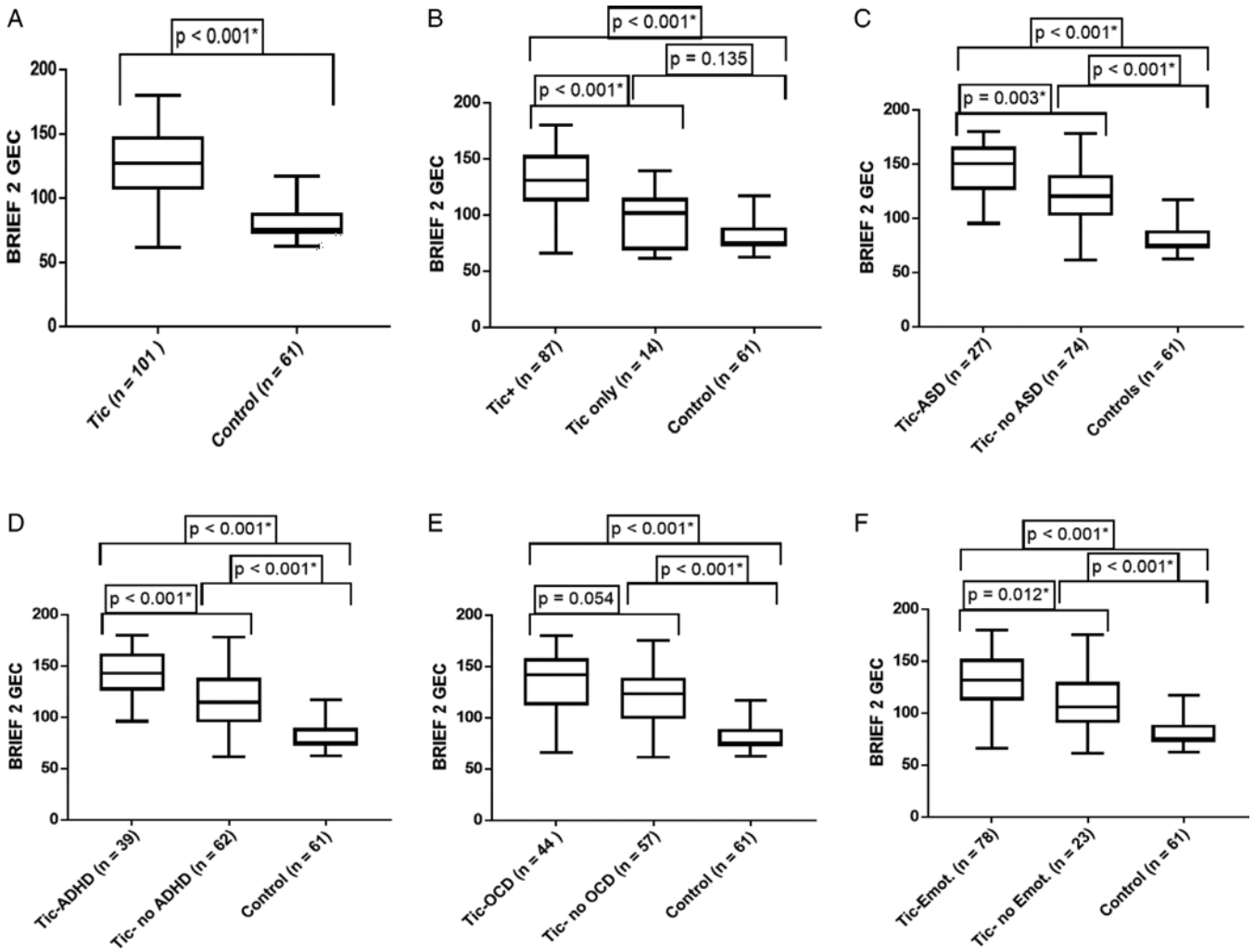

FIG. 2. Executive dysfunction associated with tic disorders and comorbidity. Note: BRIEF-2 GEC shows elevated executive function difficulties in tics participants compared to controls (A); Executive function difficulties were not present in the tics-only participants (B). The presence of all comorbidities, ASD (C), ADHD (D), OCD (E), and emotional disorder (F), was associated with higher executive function difficulties.

All BRIEF-2 subscores were elevated in the tic group compared to the controls, including the Behavior Rating Index (BRI), Emotional Regulation Index (ERI), and Cognitive Rating Index (CRI; Supporting Information Table S1; Supporting Information Fig. S2–S4).

Clinical Associations

We noted an association between increased sensory dysregulation and global executive impairment with increasing number of comorbidities as observed for the SSP2 (Fig. 3A), SPM (Fig. 3B) and BRIEF-2 (Fig. 3C) assessments. There was an increase in sensory dysregulation mean scores with an increase in number of comorbidities (SSP2, n = 161; 5.4 units per comorbidity; 95% confidence interval [CI]: 4.4–6.4; $P < 0.001$).

Similarly, there was a significant association between global executive impairment using the BRIEF-2 GEC scores with increasing number of comorbidities (Fig. 3C; SSP2, n = 161; 15.0 units per comorbidity; 95% CI: 13.0–17.1; $P < 0.001$).

As expected, significant global difficulties shown by the SDQ (Total Difficulties scores) were greatest in children with tic disorders (n = 102; median, 18; range, 0–33) compared to healthy controls (n = 61; median, 5; range, 0–16; $P < 0.001$). Higher SDQ scores were observed with a greater number of comorbidities (Fig. 3D; n = 162; 4.1 units per comorbidity; 95% CI: 3.6–4.6; $P < 0.001$).

Quality of life in children with tic disorders, measured with the PedsQL, was significantly lower than healthy controls ($P < 0.001$; Table 1). Reduced quality of life significantly reduced with the greater number of comorbidities (Fig. 3E; n = 162; –9.0 units per comorbidity; 95% CI: –10.3 to –7.7; $P < 0.001$). By contrast, tic severity (YGTSS) was not influenced by the presence of comorbidities (Fig. 3F; n = 100; 1.3 units per comorbidity; 95% CI: –1.7 to 4.3; $P = 0.384$).

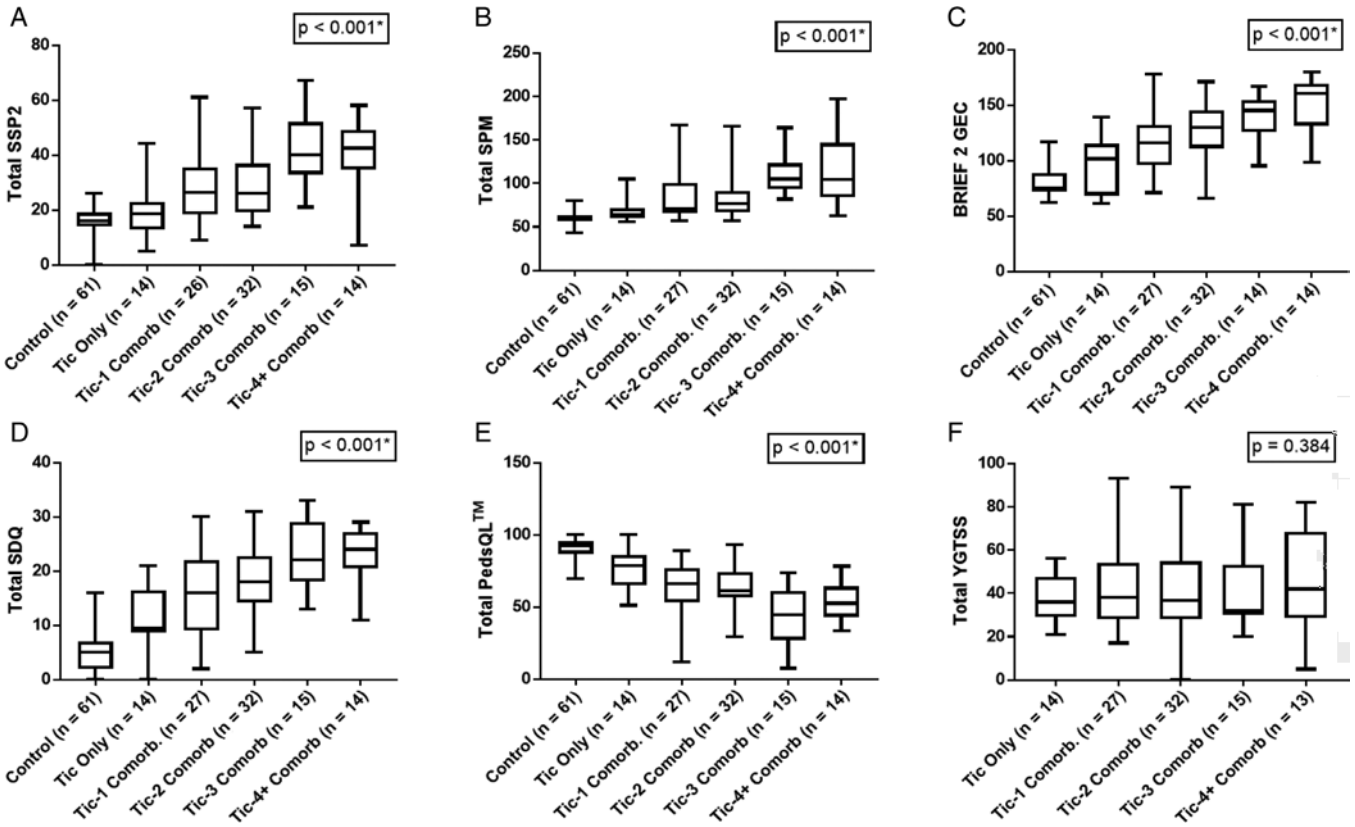

FIG. 3. Increasing impairment was associated with increasing number of comorbidities. Note: Increasing comorbidity was associated with increasing sensory dysregulation using SSP2 (A) and SPM (B), increasing executive function difficulties using BRIEF-2 GEC (C), increasing difficulties using SDQ (D), and decreasing quality of life (PedsQL) (E). By contrast, increasing comorbidity did not influence tic severity using YGTSS (F).

Within-Group Analysis for Tic Disorder Group

There was a strong positive correlation between sensory dysregulation (Total SSP2 score) and executive function difficulties (BRIEF-2 GEC score; n = 100; rho = 0.742; P = 0.001) when comparing the data of all participants with tic disorders. In participants with tic disorders, there was a strong positive correlation between sensory symptoms (Total SSP2 score) and all subscales on the BRIEF-2, including the BRIEF-2 BRI (n = 102; rho = 0.672; P = 0.001), BRIEF-2 ERI (n = 102; rho = 0.711; P = 0.001), and BRIEF-2 CRI (n = 102; rho = 0.666; P = 0.001; Supporting Information Fig. S6). There was mild positive correlation between sensory dysregulation (SSP2) and tic severity (YGTSS score; n = 100; rho = 0.214; P = 0.032) in the overall tic group. There was a strong negative correlation between sensory dysregulation (SSP2) and quality of life in the total tic cohort (PedsQL; n = 101; rho = –0.629; P < 0.001).

TABLE 1. Total median and range (brackets) scores for all assessment tools used with children with tic disorders compared to healthy controls

Assessment Tools	Tic Cohort n = 102	Controls n = 61	*P* Value
Total SSP-2	30 (5–67)	16.00 (0–26)	<0.001*
Total SPM	81.00 (56–197)	59.00 (43–80)	<0.001*
Total BRIEF-2 GEC	127.00 (61–180)	75.00 (62–117)	<0.001*
BRIEF-2 BRI	25.00 (12–36)	15.00 (12–26)	<0.001*
BRIEF-2 ERI	34.00 (16–48)	19.00 (16–43)	<0.001*
BRIEF-2 CRI	66.00 (32–96)	43.00 (32–64)	<0.001*
Total YGTSS	37.00 (0–93)	—	—
Total SDQ	18.00 (0–33)	5.00 (0–16)	<0.001*
Total PedsQL	61.41 (7.61–100)	92.39 (69.57–100)	<0.001*

*P < 0.05 was interpreted as significant for all analyses. All scores were elevated in the tic patients.

Discussion

Our primary finding was that sensory dysregulation was positively correlated with the number of comorbidities in children with tic disorders. There was increased dysregulation not only in the total sensory scores, but also in all the subscores for all sensory areas across all sensory assessment tools used for this cohort. The children with tics-only had the lowest sensory dysregulation symptoms, and this was not statistically significant when compared to healthy controls, although the number of children in this group

TABLE 2. Total median and range (brackets) scores for all sensory assessment tools comparing tic patients and controls for total and subscores of sensory symptoms

Sensory Questionnaire Results	Tic Cohort (n = 101)	Controls (n = 61)	*P* Value
Total SSP2	30.00 (5–67)	16.00 (0–26)	<0.001*
SP2			
Seeking	40.00 (4–84)	20.00 (0–37)	<0.001*
Avoiding	49.00 (4–96)	24.00 (1–52)	<0.001
Sensor	36.00 (8–91)	23.00 (1–36)	<0.001
Bystander	36.00 (4–97)	24.00 (0–42)	<0.001
Auditory	19.00 (5–40)	10.00 (1–21)	<0.001
Visual	10.00 (0–27)	7.00 (0–17)	<0.001
Tactile	17.00 (0–55)	11.00 (0–19)	<0.001
Vestibular	15.00 (0–40)	8.00 (0–19)	<0.001
Proprioception	11.00 (0–40)	8.00 (0–16)	<0.001
Oral	18.00 (0–50)	11.00 (0–20)	<0.001
Conduct	20.00 (2–40)	11.00 (1–20)	<0.001
Social emotional	35.00 (1–70)	17.00 (0–44)	<0.001
Attentional	18.00 (0–50)	11.00 (0–23)	<0.001
Total SPM score	81.00 (56–197)	59.00 (43–80)	<0.001*
Social participation	20.00 (10–37)	11.00 (10–23)	<0.001
Vision	15.00 (11–33)	11.00 (11–22)	<0.001
Hearing	11.00 (8–32)	8.00 (8–13)	<0.001
Touch	15.00 (11–42)	11.00 (11–20)	<0.001
Taste and smell	7.00 (5–19)	5.00 (5–8)	<0.001
Body awareness	15.00 (10–37)	10.00 (10–17)	<0.001
Balance and motion	15.00 (11–35)	12.00 (11–17)	<0.001
Planning and ideas	17.00 (9–36)	9.00 (9–17)	<0.001

*$P < 0.5$ was interpreted as significant for all analyses. All sensory scores including subscores were elevated in the tic patients.

was small (n = 14). The tic cohort predominantly consisted of patients with TS (80.4%). Importantly, we observed a similar rate of isolated TS without comorbidity compared with previous cohort studies, where 80% to 90% of participants had comorbidities.[49]

Although we have demonstrated that children with tics in the presence of comorbidity have increased sensory dysregulation, we are uncertain whether we are describing the same phenomenon described by Ganos and colleagues,[50] who noted that interoceptive awareness was possibly involved in the premonitory urge in adults with tics. We showed a strong positive correlation between sensory questionnaires, providing convergent validity to support existing literature in children with ASD.[51]

Following on from our own findings and the study completed by Weisman and colleagues,[7] we speculate that the sensory dysregulation experienced by children with tic disorders and comorbidities is a problem of "salience" associated with corticostriatal dysfunction, rather than attributable to a sensory deficit. Further research is required to examine this pathophysiological hypothesis further.

We showed that children with tic disorders had higher executive function difficulties compared to healthy controls. This significant difference between tic participants and healthy controls was noted in all executive function subscales, not restricted to emotional regulation as initially hypothesized.

We have described a positive correlation between sensory dysregulation severity and executive function difficulties in children with tic disorders. In treating these complex symptoms, it would be helpful to understand whether there is a causal relationship between executive function difficulties, including self-regulation and sensory dysregulation, in children with tic disorders. We hypothesize that sensory dysregulation is a dysexecutive problem. A pilot study found promising preliminary results using the Alert program, which uses sensorimotor strategies to assist self-regulation in children with tics.[52]

There was a negative correlation between sensory dysregulation severity and quality of life. This indicates that sensory symptomology should be included in the screening of children with tic disorders, and indeed perhaps all neurodevelopmental disorders, given that sensory dysregulation can impact on quality of life and may require intervention. However, we note that there are confounders, most notably the increasing rate of comorbidity with increasing sensory dysregulation.

A limitation to the study was that participants recruited were a referral cohort, rather than population based, with the potential for severity bias. It is plausible that a population-based study of tic disorders may show different rates of sensory dysregulation and executive function difficulties. However, we are most interested in applying our findings to improve treatment paradigms, and so a treatment-seeking cohort is thereby suitable.

Furthermore, given that we recruited all sequential patients with tic disorders to this study, a wide spectrum of tic disorders were included (i.e., provisional tic disorder and persistent motor or vocal tic disorder), although TS constituted 81% of the cohort. Future studies should compare the different tic diagnoses separately.

A further limitation was that a cross-sectional study design will not capture variability of tics severity in patients, given that tics wax and wane. A longitudinal study would assist in determining dynamic trends in terms of changing sensory dysregulation with the course of the tic pattern and other symptoms.

The design of our study, which recruited consecutive referred patients, led to a typically complex cohort with overlapping comorbidities. As a consequence, although we have shown that all comorbidities appear to be associated with increasing sensory dysregulation symptoms, we could not determine which comorbidities had the most influence on sensory dysregulation. Only recruiting larger cohorts with isolated comorbidity (such as tic-OCD only) will definitively address the relative contributions of each comorbidity.

Future studies should address whether the premonitory urge (using the Premonitory Urge for Tics Scale [PUTS][34]) and sensory dysregulation are associated; however, the younger participants in this study had

difficulty understanding the questions in the PUTS (data not included).

Finally, we noted limitations to the current sensory tools. The most predominant limitation was that many of the tic participants, or their parents, described sensory dysregulation not recorded by either questionnaire. These included sensitivity to "people-made" noise, including cutlery scraping on crockery, chewing of food, or the "scratching" sounds of clothing. Furthermore, although the sensory tools captured the sensory dysregulation we had commonly observed in our clinical experience of children with tics (tactile, auditory, visual, and oral), the SSP2, SP2, and SPM under-represented the size of the sensory dysregulation in a pediatric tic population, given that these sensory tools capture broader symptoms that overlap with other brain disorders.

We suggest that future research should focus on the development of a more sensitive tool to assess sensory dysregulation in children with tics and other neurodevelopmental disorders and to better target treatment effects. There is also the possibility that sensory dysregulation, premonitory urge, and tic severity are linked in terms of the sensory nature of these experiences. Therefore, a questionnaire that captures sensory dysregulation, impact on daily function, and the premonitory urge experienced in children is indicated.

Conclusion

Children with tic disorders in the presence of comorbidities experience increased sensory dysregulation and executive dysfunction and reduced quality of life compared to healthy controls. The presence of increasing number of comorbidities resulted in worsening results for all of these measures. There is a strong positive association between executive dysfunction and sensory dysregulation in children with tic disorders. Sensory regulation should be considered an executive function, and sensory dysregulation should be considered part of the neurodevelopmental spectrum observed in children with tics and comorbidity. ■

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Supporting Data

Additional Supporting Information may be found in the online version of this article at the publisher's web-site.

CHAPTER FOUR

An Exploratory Study into an Adapted use of the Alert Program® for Tic Disorder in Children

This chapter details the pilot studies conducted to trial the Alert Program® in a paediatric population, as a sensory-based approach to reduce tics. As this study was published, this chapter is presented in the published format of the paper. A preface to the chapter, as well as an authorship statement, has been provided. Thereafter the sections: abstract, background, methods, results, and discussion will follow in the format of the *Australasian Psychiatry* journal. A list of references will be found at the end of the chapter.

Preface

With no disease-specific treatment for children with tic disorders and limitations with current treatment approaches, there is a critical need for a feasible complementary assessment and treatment program for children with tic disorders (1, 4, 55, 57). Verification that children with tic disorders and comorbidities have significantly higher rates of sensory dysregulation symptoms (Study 1, Chapter 3) confirms the need to trial a sensory-based treatment approach for children with tic disorders. This study pilot tested the Alert Program®, a sensory based-approach with twelve children's tic disorders in an open-label prospective design.

The Alert Program® strategies were provided to twelve study participants by a senior paediatric occupational therapist with 16 years of experience working at the Children's Hospital at Westmead, certified and trained in the Alert Program®. The occupational therapist was trained and experienced in sensory integration assessment and intervention and had experience working clinically with children with tic disorders. Of the twelve study participants who commenced the study, ten completed the program, and two dropped out after the first session. Both of these study participants chose to leave the study due to family stressors and not being able to attend appointments.

As the study participants were experiencing tics at the commencement of the program, it was essential that the treatment prioritised the relief of tics over and above the teaching of emotional regulation concepts. Therefore sensory strategies were provided to study participants from the initial session rather than following the order of the Alert Program®, which outlines spending several sessions focusing on teaching the concept of emotional regulation and identifying participants' sensory preferences first. For this reason, the program was modified from having twelve sessions or stages, to three. The treatment sessions were provided monthly rather than

weekly, so families had time to purchase the sensory-based products (if required) and implement the strategies to determine their effectiveness before re-evaluating them at the following session. Follow-up phone calls at the end of the three sessions (T4 as per Chapter 4 Figure 1) were scheduled rather than on a needs basis.

When determining which sensory-based proxy report measure to implement for the study, the researcher selected the Child Sensory Profile 2 over the Sensory Processing Measure. From our prevalence study data (Study 1, Chapter 3), there was a strong positive correlation between the scores of both of the proxy-report sensory-based measures, the SSP2 and SPM sensory assessments ($n = 162$; $rho = 0.842$; $P < 0.001$). Therefore, it would have been appropriate to use either measure, but since the CSP2 had been used more frequently in published research studies, this was selected as the preferred sensory-based measure for this research study.

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International Conferences

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National Conferences

Soler. N, Hardwick. C, Perkes. I., Mohammad. S., Dossetor. D., Nunn, K., Bray. P, Dale R. Preliminary Investigation into sensory strategies for tic disorders in children. ***The University of Sydney Children's Hospital Westmead Clinical School Discipline of Child and Adolescent Health, Higher Degree Research Conference, Parramatta. August 2017.***

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Authorship Statement

The co-authors of the paper ‘‘An exploratory study into an adapted use of the Alert Program® for tic disorder in children.’’ confirm that ***Nicolette Soler*** has made the following contributions:

- Conception and design of the research
- Collection and extraction of all data
- Analysis and interpretation of the findings
- Drafting and revising the manuscript and critical appraisal of content

As the primary supervisor for the candidature upon which this thesis is based, I can confirm that the above authorship attribution statement is correct.

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Abstract

Objectives: This preliminary study explored whether an adapted approach to the Alert Program, that uses sensorimotor strategies, might assist with management of tic disorders in children.

The Alert Program, a program that uses sensorimotor strategies for self-regulation in children with neurodevelopmental disorders, had not been trialled with children with tic disorders.

Methods: Ten children with tic disorder were assessed using the Dunn Sensory Profile 2 (SP2), the Yale Global Tic Severity Scale (YGTSS) and the Parent Tic Questionnaire (PTQ). Participants attended three 60–90-minute appointments with an occupational therapist and clinical psychologist for implementation of the adapted Alert Program.
Results: The YGTSS showed tic reduction in all participants. The total YGTSS pre-intervention mean score of 46.5 improved to 17.7 post-therapy. Five participants reported no impairment post-therapy. PTQ scores reduced in nine participants. On the SP2, 30% of participants scored as having sensory sensitivities that impaired daily function.
Conclusions: This exploratory study found trialling an adapted approach to the Alert Program that uses sensorimotor-based approach decreased tic severity in children with tic disorders. A randomised controlled trial is needed to establish the effectiveness and feasibility of this approach.

Keywords: Tourette syndrome, tic disorders, sensory, child

Tics are repetitive, stereotypical, rapid, non-rhythmic movements or vocalisations. Tourette syndrome (TS) is diagnosed when motor and vocal tics are present for over one year. Tics begin in childhood, severity peaks in early adolescence and declines in young adulthood.[1,2] Children with TS experience increased emotional, behavioural and social difficulties, and higher rates of insecure peer attachment, compared with typically developing peers.[2]

Behaviour therapy and psychoeducation are first-line treatments for tics.[3,4] Existing behavioural therapies are time-intensive and not all children respond to treatment.[5] The premonitory urge (PU) is described as localised discomfort immediately before a tic and may contribute to tic generation.[6] The PU is generally not reported in children under 10 years of age.[7] In addition to the PU sensation, broader somatic hypersensitivity has been described in people with tics.[8] Therefore, tics seem to

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involve sensorimotor phenomena, rather than a 'pure' movement disorder as tics have prominent sensory features.

The Alert Program[9] uses sensorimotor strategies to assist with children's self-regulation and attention. This program, although used to treat children with other neurodevelopmental disorders, has not been used with children with tic disorders.

Sensorimotor-based therapy has yet to be trialled for tic management. However, these strategies have been applied to related clinical populations, for example children with autism.[10,11]

At The Children's Hospital at Westmead (CHW) Tic Clinic, clinical experience suggested a sensorimotor framework had relevance to understanding and managing tics. We suspected an increased prevalence of sensory sensitivities in children with tic disorders. Families frequently report that tics abate during physical activity such as sport. Children successful in effective suppression of their tics use self-initiated sensorimotor strategies (e.g. chewing gum, stretching and wearing tight clothing).

We initially trialled a sensorimotor approach to manage the symptoms of a child hospitalised for a cheek-biting tic. Through collaboration with a sensory trained occupational therapist, the child trialled chewing on ice cubes and frozen cloth, which reduced the tic.

This led to further trials of sensorimotor strategies resulting in symptom reduction and high treatment acceptability.

A sensorimotor-based therapy to treat tic disorders in children was developed with a view to formulating a standardised protocol for further study.

Materials and methods

Participants

Twelve participants diagnosed with tic disorder and current patients of a paediatric neurologist or child and adolescent psychiatrist working at CHW were recruited to the sensorimotor therapy program. Ten participants completed the program.

All parents consented to their child's participation and all identifying information has been removed. Ethics approval was obtained by the Sydney Children's Hospital Network, Clinical Governance Unit for a file audit (Charli number 5554). All participants received a diagnostic assessment and education on tic disorders pre-referral to the program. Participants with comorbid diagnoses were included. Table 1 shows the participant demographics (mean age 11 years 4 months, age ranged between 7 years 7 months to 16 years 7 months, 9M:1F) and comorbidity (three attention deficit hyperactivity disorder (ADHD), four generalised anxiety disorder).

Four participants were on established medication regimes, and data on concurrent use of non-pharmacological treatment was collected (Table 1). No changes in medication or other health interventions occurred during sensorimotor intervention. All participants attended mainstream education.

Assessment instruments

The YGTSS,[12] Parent Tic Questionnaire (PTQ)[13] and the Dunn Sensory Profile 2 (SP2)[14] were used on a clinical basis. All questionnaires were used at baseline, and outcomes were assessed with the YGTSS and PTQ at one month after the third therapy session.

Intervention

Therapy was provided over three appointments at monthly intervals by an occupational therapist (NS) and clinical psychologist (CH) (Figure 1). The participant and their parent(s) were present for all appointments, which lasted between 60 and 90 minutes and a home program summary was provided (individualised strategies and resource suppliers). Therapists offered between-appointment telephone and email support regarding program implementation and compliance monitoring.

Therapists

Both therapists were present for the duration of all appointments. The clinical psychologist focused on family concerns, tic severity, frequency and therapy progress. The psychologist provided a supportive stance but comprehensive behaviour intervention for tics or other forms of psychological therapy were not provided. The occupational therapist assessed sensory features, prescribed sensorimotor strategies and provided details regarding resource use and precautions.

General principles regarding intervention

Strategies were used in the home, community (e.g. at school and leisure facilities) and travelling to and from school. Sensorimotor strategies were recommended for use (i) regularly during the day, (ii) before an anticipated tic bout (i.e. end of the school day), and (iii) when the child felt a PU. Participants provided within-session verbal participant feedback to guide selection of sensorimotor strategies.

'The sensorimotor strategies (further outlined in Table 2) used by participants were categorised into the following domains.

SP2: Dunn Sensory Profile 2

1. Active body-focused movements (e.g. resistant stretch exercises)

Table 1. Demographics and clinical characteristics

Participant	*Gender*	*Age years: months*	*Primary diagnosis and comorbidities*	*Sensory symptoms on SP2*	*Concurrent pharmacotherapy*	*Concurrent non-pharmacological interventions*
1	M	9:10	Tourette syndrome Attention deficit hyperactivity disorder (ADHD)	Nil	Nil	Psychology
2	M	10:5	Tourette syndrome	Vestibular Proprioception	Nil	Chinese medicine Acupuncture
3	M	14:3	Tourette syndrome Anxiety disorder	Nil	Clonidine Citalopram	Psychiatry Physiotherapy
4	M	16:7	Chronic vocal tic disorder	Nil	Nil	Nil
5	M	7:8	Tourette syndrome	Nil	Nil	Nil
6	M	10:5	Chronic tic disorder Anxiety disorder	Vestibular Proprioception Tactile	Clonidine	Nil
7	M	11:7	Tourette syndrome ADHD Enuresis	Auditory Tactile Proprioception	Methylphenidate	Nil
8	M	14:3	Tourette syndrome ADHD Anxiety disorder	Auditory Tactile	Atomoxetine Clonidine Fluoxetine	Psychologist
9	M	7:7	Chronic tic disorder Anxiety disorder	Nil	Nil	Vitamin D supplement Multivitamins Iron supplement
10	F	12:0	Tourette syndrome	Vestibular Tactile	Nil	Osteopath Magnesium supplement

2. Calming sensory resources
3. Active resource-focused activities with either a sensorimotor or cognitive component (fidget toy or puzzle)
4. Passive body-focused activities done by the parent on the child (e.g. massage).

Session 1

Assessment of clinical features and functional impairment of the tic disorder included the participant's functioning of daily life activities and any sensory symptoms.

Therapy formulation used assessment of tic symptoms and sensory profile. This was informed by analysis of the SP2, YGTSS, PTQ, clinical observations and clinical interview (Tables 1 and 2). General education and information on self-regulation using the Alert Program terminology was used. Explanation and trial of the recommended sensorimotor strategies were implemented in the session.

Sessions 2 and 3

The participants' tics and their use and the effectiveness of implemented strategies were reviewed. Problem-solving of any application issues occurred such as adjustment of frequency and/or intensity of strategies and trialling additional strategies if required.

Follow-up

One month following the third session, progress and adherence was assessed using the YGTSS and PTQ.

Statistical comparison

A non-parametric two-tailed Wilcoxon Signed Rank Test was used to compare the pre- and post-group values for the PTQ and YGTSS.

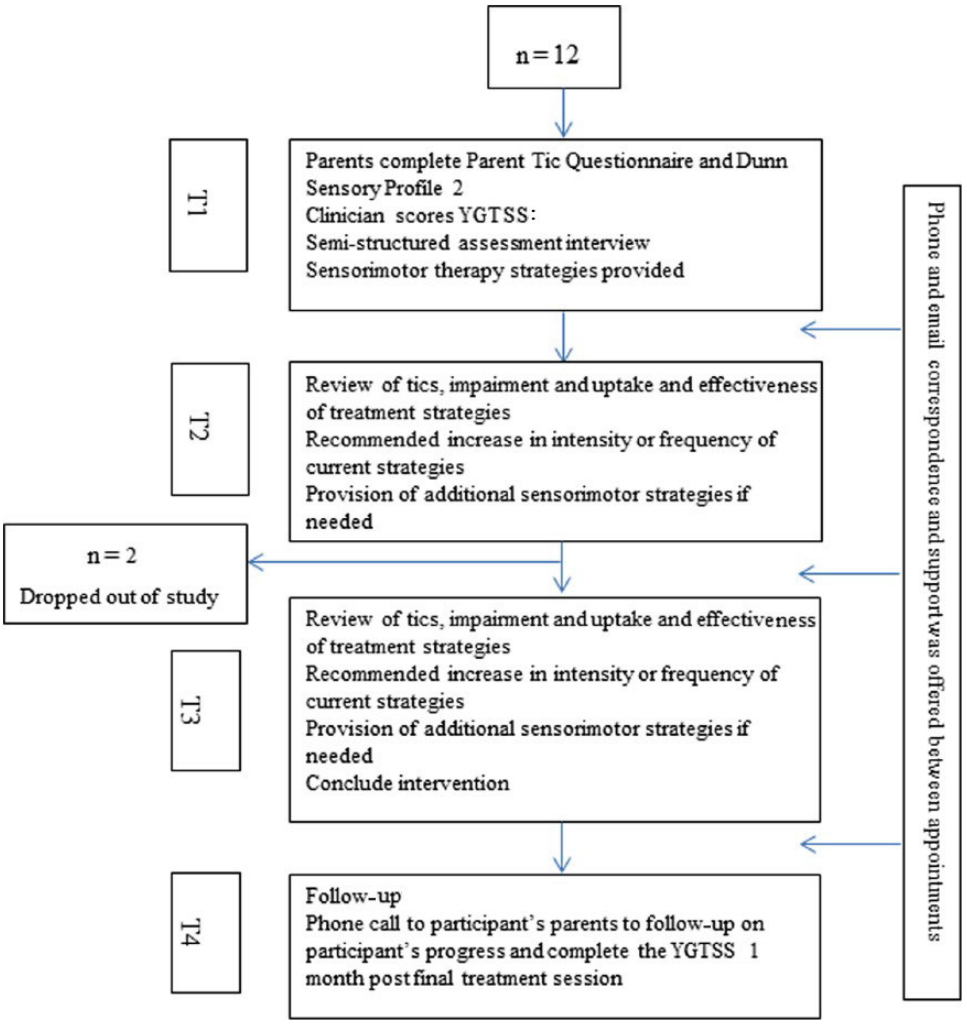

Figure 1. Flow diagram of study methodology.
YGTSS: Yale Global Tic Severity Scale

Results

Total YGTSS pre-intervention mean score of 46.5 improved to 17.7 post-therapy (Figure 2). All participants showed reduced tic severity after therapy. Five participants (nos. 1, 2, 5, 6 and 9) reported absence of any impairment after therapy. Significant sensory symptoms affecting daily function (per SP2 scoring criteria) were detected in 30% of participants.

PTQ scores reduced post-therapy in nine participants (Figure 3); however, the group change was not significant ($p = 0.16$), and one participant showed an increased post-therapy tic. For this participant, different parents rated the questionnaire, whereas the questionnaires were completed by the same parent for all other participants.

Clinical impressions

All participants showed a reduction in the intensity and/or the frequency of motor and vocal tics following therapy on the YGTSS. Most participants showed high acceptance and tolerability to the strategies trialled. Participants reported regular use of strategies at home. One barrier to implementation was a delay in accessing prescribed tools, with family stress and competing demands being advised as the reason.

Discussion

Our study applied a sensorimotor therapy paradigm to a paediatric tic disorder cohort. Our rationale acknowledged the role of the PU and broader sensory symptoms that were over-expressed in this population, and that many children and families reported tics reduced during physical activity. We therefore trialled an adapted version of the Alert Program that used sensorimotor strategies to manage tics. This approach was useful in children with and without identified sensory features. The aim was to manage tics using sensorimotor-based strategies, rather than provide therapy to treat any underlying sensory difficulties that some participants experienced.

Table 2. Summary of tic symptoms, sensory sensitivities and individualised sensorimotor strategies implemented

Participant	*Current tic type*	*Most severe current tics*	*Sensory symptoms reported by client/family*	*Pre-YGTSS score*	*Sensory strategies*	*Post-YGTSS score*
1	Motor & Vocal	Head shaking & arm movements simultaneously	Vestibular issues - difficulty sitting still, easily dizzy, motion sickness Tactile- fidgety, always busy with hands, high pain threshold Proprioception-seeking behaviour	35	Movement cushion Movement breaks Sport activities Graded whistles Joint compression Compression clothing Crunchy foods	17
2	Motor & Vocal	Self-injurious behaviour Blinking Arm flexion Facial grimacing Throat clearing Coughing	Vestibular and proprioception-seeking behaviour	55	Movement cushion Movement breaks Deep pressure program Push-ups	3
3	Motor & Vocal	Complex neck and shoulder Neck Eye squint	Problems wearing certain clothing Difficulty coping with multi-sensory environments	47	Joint compression Rock climbing Chin lifts Weighted lap blanket Gym ball	21
4	Motor & Vocal	Grunting Facial grimacing	Plays continuously with hands and fidgety †	30	Slow, controlled swallowing Drinking straw Chewing gum Fidgets toys	14
5	Motor & Vocal	Squeaks Grunting Quick gasp Head nod Eye blinking Facial grimacing Jaw slide Abdominal tensing Finger tapping Skipping movement Leg kick	Difficult to focus with loud noises † Tactile sensitivity to textures, clothing and clothing tags, decreased responses to pain Sensitive to smells	48	Movement cushion Movement breaks Weighted lap blanket Graded whistles Resistive putty with essential oils	7
6	Motor & Vocal	Eye blinking Facial movements Head, shoulder and arms Mouth stretching Shoulder shrug Holding breath Leg kick	Vestibular and proprioception-seeking behaviour Oral: food sensitivities, limited diet Auditory sensitivity: covers ears due to loud noises Visual sensitivities Tactile sensitivities to tags and clothing problems Difficulty with multi-sensory environments	63	Movement cushion Fidget toys Resistive putty with essential oils Exercise bands Graded whistles Chair push-ups	11

(Continued)

Table 2. (Continued)

Participant	Current tic type	Most severe current tics	Sensory symptoms reported by client/family	Pre-YGTSS score	Sensory strategies	Post-YGTSS score
7	Motor & Vocal	Yelling Repeating words Sniffing Eye blinking Facial movements Finger movements Cracking knuckles	Auditory sensitivity: dislikes loud noises Tactile difficulty with clothing tags and clothing, avoids tactile sensation, high pain threshold Proprioception sensitivities, seeks deep pressure through hugging others Multi-sensory environment problematic	45	Gloves Weighted lap blanket Fidget toys Resistive putty with essential oils Chewing gum Exercise bands Compression clothing Oculomotor exercises Chair push-ups	25
8	Motor & Vocal	Eye blinking Mouth movements Head nodding Grunting	Proprioception and auditory sensitivities Tactile sensitivities as clothing textures and tags are a problem Vestibular sensitivities with motion sickness reported	55	Resistive putty Chewing gym Crunchy foods Oculomotor exercises Movement breaks Exercise bands Fidgets toys	29
9	Motor & Vocal	Head & neck tic Eye blinking Mouth movement Arm movement Grunting	Oral sensitivities, gags on food, picky eater Tactile sensitivities to tags and clothing are, avoids messy play	26	Movement cushion Chewing gum Graded whistles Resistive putty Lycra Movement breaks	8
10	Motor & Vocal	Throat clearing Laughing Jaw clicking Head shaking Eye blinking Nose twitch Tensing arms Hand movement Leg jerking Tensing arms	High pain threshold Places objects in mouth	58	Continue dancing Resistive putty with essential oils for smell Exercise bands Movement cushion Crunchy foods Chewing gum	42

†The sensory symptoms reported by participants 4 and 5 could also be considered traits of ADHD but neither of these participants had any other diagnosed comorbidities. There could be an overlap of symptoms between various diagnoses.
Fidget toys were defined as tools that could be manipulated in the hand to assist with calming the participant.
Graded whistles were described as whistles that provided oral and ocular input but no auditory output and required the participant to use sustained, controlled breathing to make the whistle work.
YGTSS: Yale Global Tic Severity Scale

A sensorimotor approach with children with tic disorders may be a means of using sensorimotor strategies to assist in self-regulation, rather than managing sensory sensitivities in this population. As not all participants experienced comorbidity of sensory symptoms, this sensorimotor approach might act via multiple mechanisms including emotional regulation, stress and arousal reduction, distraction, education, and parent–child interaction. We acknowledge the full extent and mechanisms of the effects are not yet clearly understood.

The approach appears feasible, and cost and time effective. Importantly, children enjoyed the interventions and reported reduced tic severity and impairment.

A better understanding of the relationship between the prevalence of sensory symptoms, the PU and the tic disorders may assist in refining the sensorimotor approach and adapting the program further. The inclusion of the Premonitory Urge to Tic Scale[15] may aid in understanding this relationship.

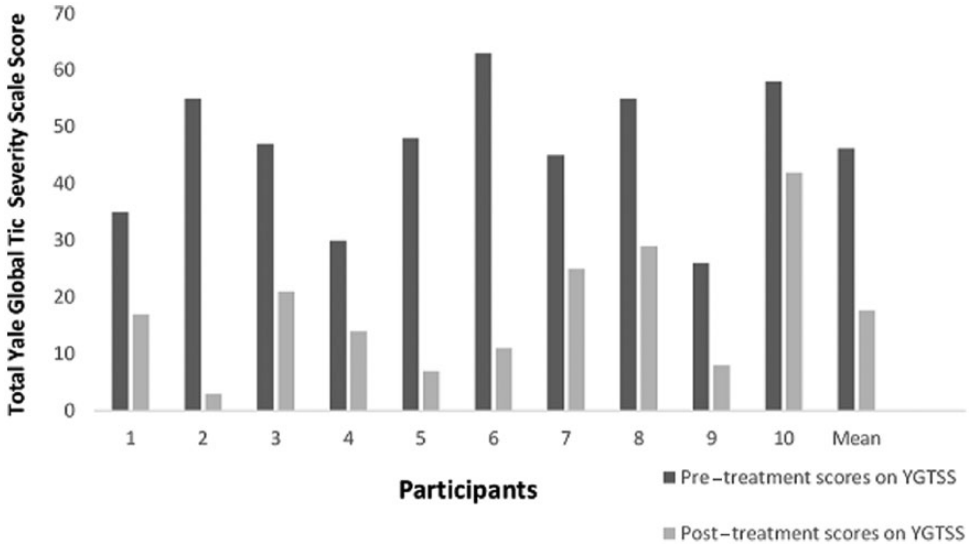

Figure 2. Total Yale Global Tic Severity Scale Score before and after treatment. The score measures the number, frequency, intensity, complexity and interference of both vocal and motor tics as well as the impairment. The higher the severity scores, the greater the impact of the tic disorder on the participant's function. The dark-grey values show the score before intervention and the light-grey values show the tic severity post-intervention. All participants show a reduction in tic severity after three appointments.

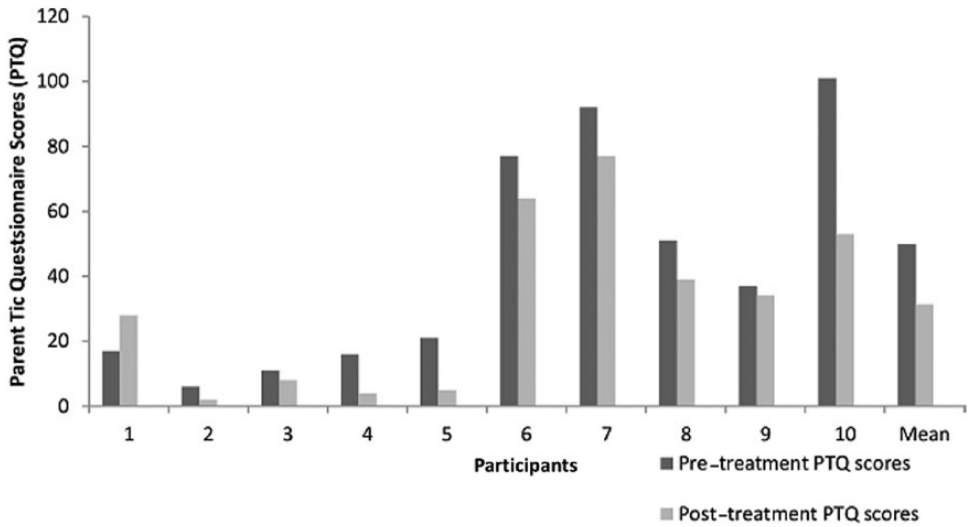

Figure 3. PTQ scores before and after treatment.

Parents were asked to rate the number and frequency of the tics their child had experienced in the past week. The results in dark grey show the scores pre-intervention and the light grey indicates results post-intervention. Participant 1 shows an increase in tic severity according to the parents post-treatment in contradiction to the reduction in tic severity as rated on the YGTSS for that participant.

The limitations of this study are inherent to a case series. Assessors and participants were unblinded, and measurement bias, case-selection bias, and the placebo effect cannot be eliminated. Participants reported daily use of the strategies, although adherence was not explicitly measured.

Given the waxing and waning nature of tics, a one-month follow-up period is too short to confirm whether the benefit is sustained. This is a limitation of this study. Further research into the long-term effects of this intervention is needed.

Whilst this study found that an adaption to the Alert Program that uses sensorimotor strategies may reduce tics, the causal mechanism was unclear. A randomised controlled trial could assess these limitations further and

assist in understanding the longer-term outcomes of the therapy.

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CHAPTER FIVE

Proxy Reported Sensory Measures Used for Children and Adolescents with Neurodevelopmental Disorders: A Systematic Review Evaluating Measurement Properties.

This chapter explains the systematic review study that was conducted to evaluate the proxy-report sensory-based measures used for children and adolescents with neurodevelopmental disorders. As this study was published, this chapter is presented in the published format of the paper. A preface to the chapter, as well as an authorship statement, has been provided. Thereafter the sections: abstract, background, methods, results, and discussion will follow in the format of the *Developmental Medicine and Child Neurology* journal. A list of references will be found at the end of the chapter.

Preface

The findings from our first two studies, the prevalence study (Study 1, Chapter 3) and the pilot study (Study 2, Chapter 4), revealed that children and their parents reported sensory dysregulation symptoms not measured on either of the commonly used sensory-based measures. Instead of further examining the Alert Program® as a treatment approach for tics, it was necessary to shift our focus to investigate if there were other proxy-report sensory-based measures that may be more suitable for use in children with tic disorders. Therefore, a systematic review was undertaken to identify all the proxy-report sensory-based measures available for children with neurodevelopmental disorders. Once we had identified all these measures, we then critically evaluated the design and development of the measures, content validity and psychometric properties of these measures. The researchers had to widen the search to include developmental disorders rather than tic disorders, as there were no sensory-based measures designed solely for this tic population. This study aimed to provide evidence in guiding the appropriate selection of a proxy-report sensory-based measure for children with tic disorders.

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Authorship Statement

The co-authors of the paper ‘‘Proxy-reported sensory measures for children and adolescents with neurodevelopmental disorders: A systematic review’’ confirm that ***Nicolette Soler*** has made the following contributions:

- Conception and design of the research
- Collection and extraction of all data
- Analysis and interpretation of the findings
- Drafting and revising the manuscript and critical appraisal of content

As the primary supervisor for the candidature upon which this thesis is based, I can confirm that the above authorship attribution statement is correct.

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SYSTEMATIC REVIEW

Proxy-reported sensory measures for children and adolescents with neurodevelopmental disorders: A systematic review

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Abstract

AIM: To determine the quality and utility of proxy-reported sensory measures for children and adolescents with neurodevelopmental disorders (such as autism spectrum disorder, attention-deficit/hyperactivity disorder, movement disorders, and intellectual disability).

METHOD: We systematically searched 11 databases. We applied the updated Consensus-based Standards for the selection of health Measurement INstruments (COSMIN) Risk of Bias checklist and criteria for good measurement properties to evaluate instrument development and psychometric properties. Findings were summarized using a COSMIN adaptation of Grading of Recommendations, Assessment, Development and Evaluations.

RESULTS: From 11 databases, 6748 articles were screened. Ninety-one full-length articles were reviewed after removing excluded studies and manual searches conducted by two reviewers. Data were extracted for 12 measures from 20 articles. Of the 12 measures, only three provided sufficient data to evaluate content validity and psychometric measurement properties. The Participation and Sensory Environment Questionnaire-Home (PSEQ-H) was the only measure that satisfied moderate content validity and moderate-to-high quality for measurement properties. These properties included: structural validity, hypothesis testing for construct validity, internal consistency, reliability, and measurement error.

INTERPRETATION: One measure, the PSEQ-H, met eight criteria for good measurement properties. To facilitate evidence-informed clinical decision-making, all psychometric properties of all 12 sensory-based, proxy-reported measures were presented. The importance of consumer engagement in measure development and the need for ongoing evaluation of measures against contemporaneous standards is recommended.

Abbreviations: COSMIN, Consensus-based Standards for the selection of health Measurement INstruments; CSP2, Child Sensory Profile 2; EPYFEI, Assessment of Sensory Processing and Executive Functions in Childhood; GRADE, Grading of Recommendations, Assessment, Development and Evaluations; PSEQ, Participation and Sensory Environment Questionnaire; PSEQ-H, Participation and Sensory Environment Questionnaire-Home; SEQ-3.0, Sensory Experiences Questionnaire-Version 3; SP2, Sensory Profile 2; SPM, Sensory Processing Measure; SPM-H, Sensory Processing Measure-Home; SPM-P, Sensory Processing Measure-Preschool; SPSRC, Sensory Processing and Self-Regulation Checklist; SSP2, Short Sensory Profile 2.

The ability to adaptively organize and regulate responses to sensory stimuli (including hearing, vision, touch, smell, taste, movement and balance [vestibular], body awareness [proprioception], and interoception) in one's environment is critical to participation in everyday activities.[1] Atypical responses to sensory stimuli are observed in behaviours incongruent to the sensation experienced.[2] Terminology used to describe these observed behaviours to sensory stimuli in children with neurodevelopmental disorders includes sensory dysregulation, sensory processing, and atypical sensory reactivity.[1,3–5] In this review, we use the term 'sensory dysregulation'.[4,6,7]

Sensory dysregulation is common in people with neurodevelopmental disorders[8–13] and is associated with impaired participation in activities of daily living.[14–18] Sensory dysregulation is a recognized diagnostic feature of autism spectrum disorder (ASD).[19] However, children with other neurodevelopmental disorders also experience sensory dysregulation. For instance, approximately 90% of children with tic disorders and other comorbid neurodevelopmental disorders experience sensory dysregulation.[20–22] Increased sensory dysregulation has also been reported in individuals with obsessive–compulsive disorder[23] and attention-deficit/hyperactivity disorder (ADHD).[24]

Sensory dysregulation is associated with decreased school participation, reduced enjoyment and engagement in daily tasks, and increased parental stress.[10,14,25,26] Accordingly, assessment and management of sensory dysregulation is an accepted part of comprehensive care for children with neurodevelopmental disorders.[27] Therapeutic approaches are commonly used to address sensory dysregulation in children with neurodevelopmental disorders, with most of these strategies having been developed for children with ASD.[5,28–31] Validated, sensitive, reliable, and responsive clinician-, teacher-, patient-, and proxy-reported outcome measures to assess treatment efficiency are necessary for clinical use in sensory dysregulation.[32]

There have been three previous systematic reviews of sensory measures.[33–35] However, two of these reviews[33,34] omitted the analysis of measure design.[36–39] Moreover, these reviews were undertaken between 2013 and 2017. Measurement evaluation methods have since progressed to incorporate criteria of measure relevance, comprehensiveness, comprehensibility, sensitivity, and fitness for purpose.[36–40] These criteria warrant consideration for existing sensory measures to improve the selection of instruments for research and clinical practice.

There is discordance in the literature about the most cited sensory outcome measures,[41] with measures often not covering the depth and breadth of patient symptoms.[20] The comprehensiveness of the Sensory Profile 2 (SP2) and Sensory Processing Measure (SPM) in children with tic disorders and comorbid neurodevelopmental conditions were brought into question because study participants reported sensory dysregulation symptoms that were not rated on either measure.[20] This brings into question the measurement design, construct, fitness for purpose, and validity of the psychometric properties of the available proxy-reported, sensory-based measures available to clinicians and researchers. Therefore, in the absence of such a review, there is a need to synthesize the available evidence to guide clinicians and researchers in selecting measures to evaluate sensory dysregulation.

What this paper adds

- Three measures provided studies on content validity and psychometric measurement properties.
- The Participation and Sensory Environment Questionnaire-Home had moderate quality for content validity studies and high-to-moderate quality evidence for psychometric properties.
- The Participation and Sensory Environment Questionnaire was the only measure that included consumer involvement through qualitative interviews and pilot testing.
- Consumer involvement in measure development is important for content validity.
- Ongoing evaluation of measures against contemporaneous standards is recommended.

This systematic review evaluates proxy-reported, sensory-based measures for children and adolescents with neurodevelopmental disorders using the Consensus-based Standards for the selection of health Measurement INstruments (COSMIN).[36–39] The complexity and volume of measures precluded appraisal of clinician- and teacher-rated sensory-based measures for children and adolescents; therefore, they are outside the scope of this systematic review.

This study was conducted between March 2020 and September 2021 and aimed to (1) identify all current proxy-reported measures relating to sensory dysregulation in children and adolescents with a neurodevelopmental disorder and (2) comprehensively evaluate the development and psychometric properties of these measures.

METHOD

The systematic review protocol was developed and registered with Prospero (CRD42020158005). COSMIN[36–39] was used to appraise the measurement properties of the proxy-reported sensory measures used with children with neurodevelopmental disorders. PRISMA 2020 standards were used to report guidelines (Appendix S1 and Table S1).[42,43]

Literature search

A search using subject heading and free text search terms relating to the population, sensory dysregulation, measures, and measurement properties was conducted on 3rd March 2020 across 11 databases. All retrieved articles were stored

in EndNote X9 (Clarivate, London, UK).[44] These databases included: Allied and Complementary Medicine Database, CINAHL, Cochrane, Complementary and Alternative Medicine, Embase, InformIT, MEDLINE, Pre-MEDLINE, PsycINFO, Scopus, and Web of Science (Tables S2 and S3). A manual search was also conducted with Google Scholar using keyword search terms, the name and abbreviations of measures, and by following the publication history of the authors of the identified measures. A manual search of the databases and websites of relevant publication companies (i.e. Acer, Pearson Clinical, Pro-Ed, Psychological Assessment Australia, Wiley, and WPS) was undertaken to ensure no measure or measurement manual was omitted (Figure S1).

Eligibility criteria

Articles were included if the study reported the development of (1) a child-, proxy/parent-, or caregiver-rated (2) multi-sensory measure (3) for children and young people aged 3 to 18 years (4) diagnosed with a neurodevelopmental disorder. The lower age of 3 years was selected because a systematic review of sensory-based measures used in infants had already been conducted.[45] Second, many measures are designed for children aged 3 years and older.[46–49] Therefore, different questionnaires would be used for children younger than 3 years.[45] The upper age of 18 years was used because the reviewers wanted to capture all assessments developed for children or adolescents.[50]

Studies reporting on participants of an age or diagnostic range broader than our inclusion criteria were included if a subgroup analysis was published or available on request. There was no limit regarding the year of publication and no restriction on publication language.

Independent reviewers (PB and NS) determined article eligibility using a two-step process (Figure S2). First, the title, keywords, and abstracts were reviewed to designate articles as duplicate, excluded, or included. Manuscripts of articles that passed this screening were then reviewed for final allocation as included or excluded. Discrepancies were resolved through discussion and consensus.

Evaluation of the quality of measurement properties

COSMIN, the accepted approach to appraise measures, was used to evaluate both the quality of studies and the quality of psychometric measurement properties of sensory-based measures through a multi-step process.[36–39] The study reviewers (PB and NS) evaluated content validity and then the psychometric measurement properties of the measures using the 10 COSMIN sequential steps.

The three sequential evaluation COSMIN processes were completed using the COSMIN methodology: (1) content validity, (2) internal structure, and (3) remaining measurement properties. Content validity is the degree to which the instrument's content represents the construct reported to be measured.[36–39] Through a measure having adequate content validity, the clinician or researcher is assured that the items on the questionnaire are relevant, comprehensive, and comprehensible regarding the construct being tested and the target population.[37] Therefore, content validity is the most important measurement property.[37] The COSMIN manual suggests that measures with high-quality evidence of inadequate content validity can be excluded from any further assessment in the systematic review.[37]

Internal structure refers to how the individual items in the measure relate to one another.[36–39] The evaluation of the remaining measurement properties mainly assesses the quality of the scale, or subscale, as a whole as opposed to each individual item on the scale.[36–39]

First, two independent reviewers (ND and PB) independently evaluated (step 1) content validity, which assesses the quality of (1) measure development and (2) content validity. The reviewers then (step 2) evaluated the internal structure of these measures, which included: (1) structural validity, (2) internal consistency, and (3) cross-cultural validity. Finally, (step 3) the following remaining measurement properties were evaluated: (1) reliability, (2) measurement error, (3) criterion validity, and (4) hypothesis testing for construct validity, which consists of convergent and discriminant validity and responsiveness (Figure S1).[36–39]

We evaluated all 12 measures in relation to all the psychometric properties (steps 2 and 3) as per our study protocol, which aimed to compare all available sensory-based measures. The COSMIN methodology suggests that only measures that score 'adequate' on content validity (step 1) should be evaluated further.[36–39,51] Many commonly used sensory-based measures would be excluded from further review.[47,51,52] Through a comprehensive evaluative approach of all measures, evidence is provided to compare clinical utility and guide the selection of measures across all psychometric properties. However, measures without evidence of content validity cannot be recommended for clinical use.

All three evaluation steps (i.e. content validity, internal structure, and measurement properties) include (1) evaluation of the methodological quality of the studies using the COSMIN Risk of Bias checklist, (2) application of criteria for good measurement properties using the COSMIN criteria, and (3) summarization[36–39] and grading the quality of evidence using the COSMIN adaptation of the Grading of Recommendations, Assessment, Development and Evaluations (GRADE) approach (Figure S1).[52,53]

The COSMIN Risk of Bias checklist was used to assess the methodological quality and screen for risk of bias in each included study to determine the trustworthiness of the reported study results.[36–39] The studies were rated on a 4-point score: very good (V), adequate (A), doubtful (D), and inadequate (I) for each standard. An overall score was determined by taking the lowest score across all items scored in each domain.[37]

To evaluate the quality of measurement properties, psychometric results published for each study were graded as

sufficient (+), insufficient (−), inconsistent (±), or indeterminate (?) using domain-specific COSMIN 'good measurement properties' criteria (Table S4).[36–39]

Then reviewers graded the pooled and summarized quality of evidence for each measurement property for each measure; an overall rating was determined using a COSMIN adaptation of GRADE[52,53] as specified by COSMIN.[36–38] The GRADE approach was developed for clinical trials but the COSMIN adaptation of GRADE outlined by COSMIN was developed for systematic reviews of patient-reported outcome measures. The quality of evidence refers to the confidence in the trustworthiness of the pooled or summarized result. The COSMIN adaptation of GRADE was applied to each property of each measure.

The quality of evidence was rated across five factors. These were: the risk of bias (i.e. the methodological quality of the studies); inconsistencies (i.e. unexplained inconsistency of results across the studies); imprecision (i.e. the total sample size of the available studies); indirectness (i.e. evidence from different populations other than the population of interest in the review); and publication bias (i.e. negative results are published less often).[36–38] For content validity, three factors were considered: risk of bias, inconsistency, and indirectness. For the internal structure and other measurement properties, all five factors were considered.

The quality of evidence was graded as high, moderate, low, or very low evidence according to the COSMIN adaptation of GRADE (Table S5).[36–38] It was always assumed that the quality of evidence was high. The COSMIN adaptation of GRADE has been implemented to downgrade the evidence by one or two levels per factor (i.e. moderate, low, or very low evidence) where concerns relating to the aforementioned factors exist in relation to the quality of evidence. When only a single study of inadequate quality of evidence existed, the evidence was downgraded by three levels (i.e. very low quality of evidence) (Table S5).[36–38]

After these steps, reviewers evaluated the feasibility of using these measures, formulated recommendations, and reported on the systematic review (Figure S1).

RESULTS

The results for the literature search and content validity (step 1) are discussed first. Thereafter, the results for each of the 10 measurement properties is addressed for internal structure (step 2) and other measurement properties (step 3).

Literature search

The literature search retrieved a total of 6748 publications across 11 databases. Duplicate articles (n = 2814) and articles not meeting the inclusion criteria (n = 3843) were removed after being screened by two independent reviewers to assess the title, keywords, and abstract. During the second step, the full text of 91 articles was reviewed, of which 82 articles were excluded, resulting in nine articles relating to eight different measures meeting the inclusion criteria. These nine articles related to the following measures: Assessment of Sensory Processing and Executive Function in Childhood (EPYFEI);[49] Knickerbocker Sensorimotor History Questionnaire;[54] Sensory Experiences Questionnaire-Version 3 (SEQ-3.0);[55] Sensory Processing and Self-Regulation Checklist (SPSRC);[56] Sensory Processing Measure-Home (SPM-H);[57] Sensory Processing Measure-Preschool (SPM-P);[58] Sensory Processing Scale Inventory;[50] and the Short Sensory Profile 2 (SSP2).[59]

A manual search yielded an additional 11 publications; eight were peer-reviewed journal articles and three were measurement manuals. These 11 publications related to four additional measures: the Participation and Sensory Environment Questionnaire-Home (PSEQ-H);[60–62] Participation and Sensory Environment Questionnaire-Community;[60,63] Sensory Behavior Questionnaire;[64] and the Child Sensory Profile 2 (CSP2) (Figure S2).[47]

The number of manually searched articles retrieved can be accounted for according to the following reasons: (1) measurement manuals (the SPM, SPM-P, and CSP2) would not be retrieved through the searched databases; (2) two publications were released after the search date;[62,65] and (3) cultural studies were published in journals not affiliated with the databases searched. All three publications pertaining to the PSEQ (Home and Community)[60,61,63] were only retrieved through manual searching.

In total, 20 publications (17 articles and three manuals) (Table S6) were included in this study pertaining to 12 different sensory-based measures (Table S7). All measures retrieved were proxy-reported; no child-reported questionnaires were identified. Fourteen measures were excluded due to one of the following reasons: (1) the measure was superseded by either updated versions of the same measure or by the development of a new measure (n = 5);[48,66–70] (2) because there were no publications relating to either the development or psychometrics of the measure (n = 6); or (3) the age range of the target population the measure was designed for or the psychometric studies relating to the measure were outside the scope of this systematic review and subgroup analysis was not possible (n = 3)[71–73] although data were requested (Table S8).

Many of the included studies were developed for children diagnosed with ASD. Twenty-three per cent of publications were studies involving typically developing children and no clinical sample was included. Three were studies conducted with children receiving occupational therapy interventions and three engaged children with a range of neurodevelopmental disorders. Twelve studies reported on study samples greater than 100. Eight studies recruited fewer than 100 study participants, ranging from 20 to 70 study participants. Half of the included measures were published within a 3-year period from 2017 to 2019. Five of the included studies were cross-cultural studies whereby the measures had been translated into another language (the SPM-Hong Kong Chinese version,[74] SPM-Malay version,[65] the CSP2 Spanish

version,[47] and the SSP2 Polish version).[59] One study used the English version of a measure in a cross-cultural study (i.e. the SPM-P administered to English-speaking Saudi participants).[75]

Evaluation of the measurement properties for content validity (step 1)

Of the 12 reviewed measures, three had an associated peer-reviewed published report of measure development and content validity. Those three measures were the EPYFEI,[49] PSEQ (which relates to both the home and community scales of this measure),[60] and the CSP2.[47]

For these three measures, the conceptual framework to define the construct being measured was well described for both the PSEQ and the CSP2. Although all three measures consulted with professionals in item generation and measure development, only the PSEQ (Home and Community) included patient involvement in measure design.

Patient involvement consisted of semi-structured interviews with 34 parents/caregivers; 35 items were generated. For this reason, the PSEQ (Home and Community) scored adequately for the quality of measure development, whereas the EPYFEI and CSP2 scored inadequately (Table S9). The developers of the CSP2 tested the measure to ensure grade 6 reading ability of the measure using the Flesch–Kincaid Grade Level index. However, the comprehensibility of both the CSP2 and EPYFEI measures was not tested with patients.[47,49]

Of the three measures, the PSEQ was the only measure to have moderate quality of evidence for 'sufficient' (+) overall content validity. The PSEQ was also the only measure that ensured comprehensibility. There was moderate quality of evidence for sufficient relevance and comprehensiveness and high quality of evidence for comprehensibility for the PSEQ (Table 1).

The graded evidence for both the EPYFEI and CSP2 was low (Table 1) as they scored within an inadequate range for the COSMIN Risk of Bias checklist (Table S9). There were also inconsistencies with scores in terms of criteria for quality of evidence for overall measure development, content validity, and rating of reviewer scores (Table 1).

Results for the psychometric properties of measures for internal structure (step 2)

To determine the methodological quality of all 12 measures, data were extracted and evaluated for all but one publication.[60] This single study[60] reported only on measure development and not psychometric properties.

The methodological quality ratings of the studies using the COSMIN Risk of Bias checklist is reported in Table 2. Table 3 summarizes the quality of the psychometric properties of the studies pertaining to the 12 measures based on the COSMIN quality criteria[37] (Table S3) and provides an overall

TABLE 1 Content validity ratings for relevance, comprehensiveness, and comprehensibility and quality of evidence using the COSMIN adaptation of GRADE for the three measures where content validity could be evaluated

	EPYFEI[49]					PSEQ[60,61]					CSP2[47]				
Measurement rating	Measure development	Content validity study	Reviewer rating	Overall rating	Level of evidence	Measure development	Content validity study	Reviewer rating	Overall rating	Level of evidence	Measure development	Content validity study	Reviewer rating	Overall rating	Level of evidence
Relevance rating	±	?	?	±	Low	+	?	+	+	Moderate	±	?	?	±	Low
Comprehensiveness rating	?	?	?	?	Low	+	?	+	+	Moderate	?	?	+	+	Low
Comprehensibility rating	?	?	+	+	Low	+	+	+	+	High	?	–	+	±	Low
Overall content validity rating	?	?	?	?	Low	+	?	+	+	Moderate	?	?	+	+	Low

Ratings are provided for (1) measurement development, (2) the content validity study of specific measures, and (3) the reviewer' s own rating of the content of the measures. These ratings were then synthesized into an overall rating score. COSMIN quality rating: the results are rated as either sufficient (+), whereby good measurement properties are met, insufficient (–), inconsistent (±), or indeterminate (?) when the reviewers were unable to rate the quality of evidence due to inadequate information.[39] Reviewer ratings are based on the following criteria: +, ≥85% of the items of the measure (or subscale) fulfilled the criterion; –, <85% of the items of the measure (or subscale) fulfilled the criteria; ?, no or not enough information available or quality of (part of) the study was inadequate. Quality of evidence using the COSMIN adaptation of GRADE: high evidence: there are multiple studies of at least adequate quality or there is one study of very good quality available; moderate evidence: there are multiple studies of doubtful quality available or there is only one study of adequate quality; low evidence: there is only one study of inadequate quality or there is only one study of doubtful quality available; very low: there is only one study of inadequate quality available.[36–39] Abbreviations: COSMIN, Consensus-based Standards for the selection of health Measurement Instruments; CSP2, Child Sensory Profile 2; EPYFEI, Assessment of Sensory Processing and Executive Function in Childhood; GRADE, Grading of Recommendations, Assessment, Development and Evaluations; PSEQ, Participation and Sensory Environment Questionnaire.

TABLE 2 The COSMIN Risk of Bias ratings for each of the studies for the 12 different measures across all psychometric measurement properties

Measure	Study	Structural validity	Internal consistency	Cross-cultural validity	Reliability	Measurement error	Criterion validity	Hypothesis testing for construct validity	
								Convergent validity	Discriminant validity
EPYFEI	Romero-Ayuso et al.[49]	A	V	NR	V	NR	V	V	I
CSP2	Dunn[47]	NR	V	I	D	D	V	A	A
KSHQ	Carrasco[54]	NR	I	NR	NR	NR	NR	NR	NR
PSEQ-H	Pfeiffer et al.[60,61]	NR	V	NR	A	NR	NR	NR	V
	Bevans et al.[62]	V	V	NR	A	A	V	V	A
PSEQ-C	Pfeiffer et al.[63]	NR	V	NR	D	NR	NR	NR	D
SBQ	Neil et al.[64]	NR	D	NR	NR	NR	V	V	V
SEQ-3 0	Ausderau et al.[55]	V	NR	NR	NR	NR	NR	NR	A
SPM	Parham and Ecker[57]	A	V	NR	D	A	V	A	V
	Dugas et al.[78]	NR	NR	NR	NR	NR	NR	V	NR
	Brown et al.[76]	NR	NR	NR	NR	NR	NR	V	NR
	Brown et al.[79]	NR	V	NR	D	NR	NR	V	NR
	Lai et al.[74]	NR	NR	A	NR	NR	NR	NR	NR
	Ahmad et al.[65]	NR	NR	I	NR	NR	NR	NR	NR
SPM-P	Miller Kuhaneck et al.[58]	A	V	NR	D	A	V	A	V
	Alkhalifah[75]	NR	NR	I	NR	NR	NR	NR	NR
Sensory Processing Scale Inventory	Schoen et al.[50]	A	V	NR	NR	NR	NR	NR	A
SPSRC	Lai et al.[56]	A	V	NR	A	NR	NR	A	A
SSP2	Dunn[47]	NR[a]	V	NR	D[a]	D[a]	V	A[a]	A[a]
	Chojnicka and Pisula[59]	NR	NR	V	NR	NR	NR	NR	NR

[a]Same data and sample reported on for the CSP2 and the SSP2. Two studies reporting on the psychometric properties of the PSEQ-H scale used the same data and sample.[55,56] The results report on the methodological quality of the measures using the COSMIN Risk of Bias checklist. A 4-point rating scale determines quality: very good (V), adequate (A), doubtful (D), or inadequate (I). An overall score is determined by taking the lowest score across all items scored in each domain.[36–39] Where no data were available or reported, the study was scored as not rated (NR). Abbreviations: CSP2, Child Sensory Profile 2; COSMIN, Consensus-based Standards for the selection of health Measurement INstruments; EPYFEI, Assessment of Sensory Processing and Executive Function in Childhood; KSHQ, Knickerbocker Sensorimotor History Questionnaire; PSEQ-C, Participation and Sensory Environment Questionnaire-Community; PSEQ-H, Participation and Sensory Environment Questionnaire-Home; SBQ, Sensory Behavior Questionnaire; SEQ-3.0, Sensory Experiences Questionnaire-Version 3; SPM, Sensory Processing Measure; SPM-P, Sensory Processing Measure-Preschool; SPSRC, Sensory Processing and Self-Regulation Checklist; SSP2, Short Sensory Profile 2.

psychometric quality rating for each psychometric property using the COSMIN adaptation of GRADE (Table S5).

Structural validity

Of the seven measures that had studies reporting on structural validity (EPYFEI, PSEQ-H, SEQ-3.0, SPSRC, SPM, SPM-P, Sensory Processing Scale Inventory), only three measures had a high level of evidence for sufficient quality of evidence for this measurement property (EPYFEI, PSEQ-H, and SEQ-3.0) (Table 3). Of the measures that conducted content validity studies, the EPYFEI had one study of adequate quality[49] (Table 2); therefore, there was moderate quality of evidence for indeterminate structural validity (factor loadings for items = 0.487–0.800). The PSEQ-H had one study of very good quality and no inconsistencies[62] (Table 2), which resulted in high quality of evidence for sufficient structural validity (confirmatory factor analysis scores = 0.71–0.91) (Table 3). The SEQ-3.0 had one study of very good quality[55] (Table 2); therefore, it had high quality of evidence for structural validity (Table 3). The other four measures had moderate evidence for sufficient quality of evidence (Sensory Processing Scale Inventory), insufficient quality of evidence (SPM, SPM-P), or indeterminate quality of evidence (SPSRC). The sample sizes in these studies ranged between 407 and 1732 (Table 3).

Internal consistency

Thirteen of the studies in this review[41,47,49,50,54,56–58,61–64,76] reported on the internal consistency rating for 11 of the 12 measures (SEQ-3.0 excluded), indicating that internal consistency is the measurement property most commonly reported. Of the 11 measures, nine (EPYFEI, CSP2, PSEQ-H, Participation and Sensory Environment Questionnaire-Community Scales, SPM, SPM-P, Sensory Processing Scale Inventory, SPSRC, and SSP2) had studies of very good quality. Therefore, all of these measures had high quality evidence for sufficient internal consistency (Tables 2, 3, and S10). Apart from the PSEQ-H, all measures only had one study reporting on internal consistency for each measure. The two studies reporting on the PSEQ-H[61,62] reported on the same study sample; thus, when pooling the summary of the results, the reviewers did not double the study sample. Therefore, since there were only single studies for each measure, the summary of pooled results can be found in Table S6.

Cross-cultural validity

Five studies addressed cross-cultural validity. Both the Polish version of the SSP2 (n = 1230)[59] and the SPM-Hong Kong Chinese version (n = 642)[74] had adequate methodological quality in terms of the process of translation and sample size for pilot testing (Table 2). There was a sufficient quality of evidence (one study of very good quality)[59] for the cross-cultural validity of the Polish version of the SSP2.

The SPM had low quality of evidence for inconsistent cross-cultural validity because one study had adequate quality[74] and one study, the Malay version of the SPM,[65] had inadequate quality as the sample size in each study was 30.

The methodological quality of the SPM-P administered to the English-speaking Saudi participants[75] (n = 56) and the CSP2 translated into Spanish[47] (n = 67) were inadequate because the study sample sizes were under the recommended COSMIN criteria (i.e. n = 100) (Table 2).

Psychometric properties of measures for other measurement properties (step 3)

Reliability

Only one study addressed the interrater reliability of a measure,[76] whereas the other reliability studies addressed the test–retest reliability of measures. The test–retest period for all studies was between 2 and 3 weeks, except for the CSP2, with 7 to 121 days between retest periods. No study mentioned if study participants were stable during the test–retest period. However, reviewers assumed that they were stable across all studies due to the target population being either typically developing or consisting of children with neurodevelopmental disorders in the community. One of the eight measures had high quality of evidence for reliability using the COSMIN adaptation of GRADE. The EPYFEI had high quality of evidence for sufficient reliability with one study of very good quality,[49] a sample size of 1394, and intraclass correlation coefficient scores between 0.75 and 0.93 (Table 4). The PSEQ-H had moderate quality of evidence of insufficient reliability due to inconsistencies between two studies of very good quality,[61,62] resulting in the quality of evidence being downgraded by one level. For the summary of pooled results for the PSEQ-H, intraclass correlation coefficient scores ranged between 0.5 and 0.75. Because there were scores below 0.7, the study results were insufficient. There was one study of adequate quality[56] for the reliability of the SPSRC. This study reported intraclass correlation coefficient scores of 0.91 (emotional regulation), 0.95 (sensory processing), and 0.94 for the overall score (Table 3).

Measurement error

Only five of the studies addressed measurement error. All the studies reporting on measurement error except for the CSP2 had adequate methodological quality (Table 2). The difference in time frame length between the test and retest period (7 and 121 days) resulted in a doubtful rating for this measure (Table 2). Three of the measures (PSEQ-H, SPM, and SPM-P) had moderate quality of evidence for sufficient measurement error and all measures had one study of adequate quality for the quality of the measurement properties

TABLE 3 The overall quality score per psychometric measurement property for the 12 measures is reported for (1) overall quality of ratings and (2) for the synthesis and grading of the overall quality of evidence based on the COSMIN adaptation of GRADE[37–40]

	Structural validity		Internal consistency		Cross-cultural validity		Reliability	
Measure	Overall rating[a]	Level of evidence	Overall rating	Level of evidence	Overall rating	Level of evidence	Overall rating	Level of evidence
EPYFEI	?	Moderate	+	High	NR	NR	+	High
CSP2	NR	NR	+	High	−	Very low	+	Low
KSHQ	NR	NR	−	Very low	NR	NR	NR	NR
PSEQ-H	+	High	+	High	NR	NR	−	Moderate
PSEQ-C	NR	NR	+	High	NR	NR	−	Low
SBQ	NR	NR	+	Low	NR	NR	NR	NR
SEQ-3 0	+	High	NR	NR	NR	NR	NR	NR
SPM	−	Moderate	+	High	±	Low	?	Low
SPM-P	−	Moderate	+	High	−	Very low	?	Low
Sensory Processing Scale Inventory	+	Moderate	+	High	NR	NR	NR	NR
SPSRC	?	Moderate	+	High	NR	NR	+	Moderate
SSP2	NR	NR[b]	+	High	+	High	+	Low[b]

Abbreviations: CSP2, Child Sensory Profile 2; COSMIN, Consensus-based Standards for the selection of health Measurement INstruments; EPYFEI, Assessment of Sensory Processing and Executive Function in Childhood; KSHQ, Knickerbocker Sensorimotor History Questionnaire; PSEQ-C, Participation and Sensory Environment Questionnaire-Community; PSEQ-H, Participation and Sensory Environment Questionnaire-Home; SBQ, Sensory Behavior Questionnaire; SEQ-3.0, Sensory Experiences Questionnaire version 3; SPM, Sensory Processing Measure; SPM-P, Sensory Processing Measure-Preschool; SPSRC, Sensory Processing and Self-Regulation Checklist; SSP2, Short Sensory Profile 2.

[a]Overall rating using COSMIN quality rating: quality criteria ratings.

[b]These ratings for the SSP2 are the same as the CSP2 since the same data and information were provided in the manual for both measures. The results are rated as either sufficient (+), whereby the good measurement properties are met, insufficient (−), inconsistent (±), or indeterminate (?) when the reviewers were unable to rate the quality of evidence due to inadequate information.[39] Quality of evidence based on the COSMIN adaptation of GRADE:[36–39] high evidence: there are multiple studies of at least adequate quality or there is one study of very good quality available; moderate evidence: there are multiple studies of doubtful quality available or there is only one study of adequate quality; low evidence: there are multiple studies of inadequate quality or there is only one study of doubtful quality available; very low: there was only one study of inadequate quality available. Where the study reviewers were not able to retrieve data on the psychometric properties of a measure, not reported (NR) was used.

Abbreviations: CSP2, Child Sensory Profile 2; COSMIN, Consensus-based Standards for the selection of health Measurement INstruments; EPYFEI, Assessment of Sensory Processing and Executive Function in Childhood; KSHQ, Knickerbocker Sensorimotor History Questionnaire; PSEQ-C, Participation and Sensory Environment Questionnaire-Community; PSEQ-H, Participation and Sensory Environment Questionnaire-Home; SBQ, Sensory Behavior Questionnaire; SEQ-3.0, Sensory Experiences Questionnaire version 3; SPM, Sensory Processing Measure; SPM-P, Sensory Processing Measure-Preschool; SPSRC, Sensory Processing and Self-Regulation Checklist; SSP2, Short Sensory Profile 2.

(Tables 2 and S10). No measure had high overall quality when the COSMIN adaptation of GRADE was applied. Although three measures scored within moderate quality of evidence for sufficient measurement error (PSEQ-H, SPM, and SPM-P), only the PSEQ-H reported on content validity; therefore, it is the only measure that should be considered by clinicians and researchers when selecting a measure with regard to this measurement property.

Criterion validity

All three measures that had studies on content validity (EPYFEI, PSEQ-H, and CSP2) had a single study of very good quality, resulting in high quality of evidence using the COSMIN adaptation of GRADE. The EPYFEI study[49] was downgraded one level from high to a moderate level of evidence because of inconsistencies. The EPYFEI was evaluated as having moderate quality of evidence for insufficient criterion validity. The insufficient rating came from the correlation between the SSP2 and EPYFEI ranging between 0.02 and 0.80; therefore, the results were below the expected score of 0.70 to be regarded as sufficient. The correlation between the PSEQ-H and Caregiver Strain Questionnaire was 0.7, resulting in this measure having sufficient criterion validity. The CSP2 was correlated with the Sensory Profile (0.47–0.86), Behavior Assessment for Children, Second Edition (0.28–0.82), and the Social Skills Improvement Rating Scales (−0.10 to −0.38). These pooled results meant that the overall rating for the CSP2 was indeterminate. Although the SPM and SSP2 both had high quality of evidence for sufficient criterion validity, these measures did not report on content validity; therefore, they should be used at the discretion of the clinician or researcher.

Measurement error		Criterion validity		Hypothesis testing for construct validity			
				Convergent validity		Discriminant validity	
Overall rating	Level of evidence	Overall rating	Level of evidence	Overall rating	Level of evidence	Overall rating	Level of evidence
NR	NR	–	Moderate	–	Very low	–	Very low
+	Low	?	High	+	Moderate	+	Moderate
NR	NR	NR	NR	NR	NR	NR	NR
+	Moderate	+	High	+	High	+	High
NR	NR	NR	NR	NR	NR	+	High
NR	NR	+	Moderate	+	High	+	High
NR	NR	NR	NR	NR	NR	+	Moderate
+	Moderate	+	High	±	Moderate	+	Moderate
+	Moderate	–	Moderate	+	Moderate	+	Moderate
NR	NR	NR	NR	NR	NR	?	Moderate
NR	NR	NR	NR	+	Moderate	+	Moderate
+	Low[b]	+	High	+	Moderate[b]	+	Moderate[b]

The quality of evidence for this measurement property for the SPM-H (sufficient) and SPM-P (insufficient) was not the same for criterion validity, although these two measures performed the same across all other psychometric properties (Table 2). The correlation between the SPM-H and SSP[48] was 0.72. In contrast, the same correlation between the SPM-P and SSP2 resulted in a correlation of 0.62, below the required quality criterion of 0.7.

Hypothesis testing for construct validity: convergent validity

All the convergent validity studies either used the CSP2 or SSP2[48] to compare their sensory measures against, except in two studies. Bevan et al.[62] correlated the PSEQ-H scores against the Caregiver Strain Questionnaire.[77] The PSEQ-H focuses on participation and assesses parent perspectives concerning the impact of the sensory environment on participation in daily activities for young children with ASD rather than sensory integration. Therefore, researchers used a measure other than a sensory measure as a criterion standard comparator measure to determine convergent validity. The EPYFEI[49] was the only study to use the updated version of the SSP2 rather than the original version of the measure in the study.

The convergent validity studies ranged between very good (EPYFEI,[49] PSEQ-H,[62] Sensory Behavior Questionnaire,[64] SPM[76,78,79]) and adequate (SPSRC,[56] SPM,[57] CSP2,[47] SSP2[47]) for the methodological quality of these studies (Tables 2 and S10). With this said, when using the COSMIN adaptation of GRADE to determine the overall quality of evidence for these measures, the PSEQ-H and Sensory Behavior Questionnaire both had high-quality evidence for sufficient convergent validity (Table 3).

TABLE 4 Feasibility of the three measures evaluated for content validity

Feasibility aspects	EPYFEI[49]	PSEQ[60,61]	CSP2[47]
Patient's comprehensibility	Patient's comprehensibility not tested	Pilot-tested comprehensibility through pilot testing with patients	Grade 6 reading ability of measure using the Flesch–Kincaid Grade Level index
Clinician's comprehensibility	Consulted with five occupational therapists and neuropsychologists	Content experts developed and reviewed an initial set of items for the tool	Consulted with six occupational therapists
Type and ease of administration	Completed by parent/caregiver	Completed by parent/caregiver	Completed by parent/caregiver
Length of the instrument	34 items	15 items	86 items
Completion time	15 minutes	Completion time not stated	15–20 minutes to complete
Patient's required mental and physical ability level	Reading and writing ability required	Reading and writing ability required	Reading and writing ability required
Ease of standardization	5-point Likert scale to score	5-point Likert scale to score	5-point Likert scale to score
Ease of score calculation	Manual scoring	Manual scoring	Manual scoring/can purchase Q Global for computerized administration, scoring, and reporting
Copyright	Not stated	Beth Pfeiffer[60–62]	PsychCorp, Pearson Clinical Assessment
Cost of an instrument	No cost	No cost	• SP2 Administration Manual: A$158.00 • SP2 Child Record Form 3:00–14:11 (25 pack) A$115.00 • Q Global Sensory Profile 2: Unlimited use scoring 1-year subscription: A$45
Required equipment	Writing implement and EPYFEI questionnaire	Writing implement and PSEQ questionnaire	Writing implement and SP2 questionnaire If uses Q Global, will need access to computer and Internet
Availability in different settings	Measure can be completed in different settings	Measure can be completed in different settings PSEQ-H and PSEQ-C measures Teacher questionnaire available[a]	Measure can be completed in different settings School companion questionnaire available[a]
Regulatory agency's requirement for approval	Not stated	Clinician	Speech pathologist, allied health, special education and human resources professionals, medical practitioner

[a]School/teacher versions of measures are outside the scope of this systematic review; they have been listed but their psychometric properties have not been evaluated.
Abbreviations: CSP2, Child Sensory Profile 2; EPYFEI, Assessment of Sensory Processing and Executive Function in Childhood; PSEQ, Participation and Sensory Environment Questionnaire; PSEQ-C, Participation and Sensory Environment Questionnaire-Community; PSEQ-H, Participation and Sensory Environment Questionnaire-Home; SP2, Sensory Profile 2.

The EPYFEI had a very low quality of evidence because the single hypothesis was not confirmed in the study.[49] The EPYFEI was hypothesized to correlate most strongly with the sensory processing scale of the SSP2 and least strongly with the behaviour scale of the SSP2. This was not proven since the total score obtained for the EPYFEI had a high positive correlation with the SSP2 behavioural subscale (0.80, $p < 0.001$) and the SSP2 sensory subscale (0.68, $p = 0.008$).

Discriminant validity

Twelve studies reported on discriminant validity for the different measures. Subgroup analysis was conducted between children with ASD (including Asperger syndrome, ASD, and pervasive developmental disorder-not otherwise specified) and neurotypical children. The study for the EPYFEI reported on a clinical sample that included study participants with various neurodevelopmental disorders (i.e. ADHD [$n = 95$, 5.5%]; ASD [$n = 84$, 4.8%]; language-specific disorders [$n = 106$, 6.1%]; developmental delay [$n = 15$, 0.9%]; and other neurodevelopmental disorders [$n = 83$, 4.8%]). For both the SPM and SPM-P parent questionnaires, the comparator group consisted of children receiving occupational therapy.

Although 10 different subgroups were reported on for discriminant validity for the CSP2, and it scored adequately for the quality of discriminate validity, each of these groups had small sample sizes (i.e. developmental delay [$n = 11$]; ASD [$n = 78$]; ADHD [$n = 96$]; dual diagnosis of ASD and ADHD [$n = 24$]; learning disability [$n = 45$]; gifted and talented [$n = 18$]; intellectual disability [$n = 9$]; Down syndrome [$n = 9$]; English as a second language [$n = 7$]; and other [$n = 62$]).

For the Sensory Processing Scale Inventory, the statistical method used to determine discriminant validity between subgroups was appropriate for hypothesis testing. Researchers attempted to match the typically developing cohort with the clinical sample.[50] However, there was still a significant difference in age ($z = 5.25$, $p < 0.1$), with typically

developing participants being slightly older (mean = 8 years 2 months, SD = 2 years 5 months) than the clinical sample (mean = 6 years 8 months, SD = 2 years 5 months). Although the effect size was small, the two groups differed in sex distribution, with proportionally more males in the clinical sample than in the typically developing group ($\varphi = 0.15$). Ethnicity and socioeconomical statistical data were also not reported.[50]

Responsiveness

To determine the responsiveness of the measures, the term 'responsiveness' was included in the search strategy, yet none of the studies reported on this measurement property for any measure. The SP2, SSP2, and EPYFEI were all screening measures and not used as pre-/post-test measures; therefore, it is not appropriate for these measures to report on responsiveness.

Feasibility

Information on the feasibility of implementing the three measures that provided information on content validity is provided in Table 4. All three measures can be feasibly implemented by clinicians and researchers. The measures vary in the number of items (the EPYFEI has 34 items, the PSEQ has 15, and the SP2 has 86) and cost (the EPYFEI and PSEQ are freely available, the SP2 requires the user to purchase the administration manual and record forms). All three measures use a 5-point Likert scale.

DISCUSSION

To our knowledge, this is the first systematic review evaluating multiple sensory dysregulation measures for children and adolescents using current best practice measurement standards according to COSMIN.

Twelve measures were assessed across 20 publications that provided validation data; they included three manuals. Of the 12 measures reviewed, only three (EPYFEI, PSEQ, CSP2) provided information on the development of the measure and content validity. Although the EPYFEI, PSEQ, and CSP2 were all designed through cooperation with professional experts, only the PSEQ included consumer involvement through qualitative interviews and pilot testing. This is despite the essential nature of cooperation and consumer engagement in developing items that constitute a measure to ensure relevance, comprehensiveness, and comprehensibility to the patients completing them.[36–39]

The other nine measures did not describe measurement design, nor were content validity studies conducted. In the absence of evidence that the measures are relevant, comprehensive, and comprehensible, clinicians and researchers should question the usefulness of these measures.[36–39]

When studies reporting on the PSEQ-H are evaluated using the COSMIN methodology, it is the most comprehensive, comprehensible, relevant, and psychometrically robust measure of the 12 measures evaluated and is recommended for children aged 2 and 7 years. For children older than 7 years, two measures reported on measure development and content validity, that is, the EPYFEI (designed for Spanish children aged 3–11 years) and the CSP2 (intended for children aged 3–14 years 11 months). Although content validity studies were provided for the EPYFEI and CSP2 measures, the quality of evidence was low for relevance, comprehensiveness, and comprehensibility. Of these two measures, the EPYFEI had the better quality of evidence across the psychometric measurement properties, but cross-cultural studies must be conducted to use this measure with an English-speaking population. The SEQ-3.0 (for ages 2–12 years), SPM-H (5–12 years), and Sensory Processing Scale Inventory (4–18 years) were designed to be used with an older age range. However, no measure development and content validity studies have been published for these measures; therefore, they should be used at the clinician's discretion.

Seven measures (EPYFEI, PSEQ-H, SEQ-3.0, SPM, SPM-P, Sensory Processing Scale Inventory, SPSRC) had evidence of structural validity. To determine structural validity, newer psychometric methods, such as item response theory and Rasch modelling, are recommended;[80,81] however, uptake of these methods across the studies was limited. Reasons for this include its computational complexity and limited availability of user-friendly analytical software.[82] Of the discriminant validity studies, nine were conducted with children with ASD. Understandably, the focus has been on testing these measures with children with ASD because of the high prevalence of sensory dysregulation in this cohort.[19] However, there needs to be a focus on developing measures for a broad range of neurodevelopmental disorders.[20–24]

Measures from the same suite of tools, such as the SPM and the SPM-P (used with different age ranges), PSEQ-H, Participation and Sensory Environment Questionnaire-Community Scales (used for different environmental settings), and the CSP2 and SSP2 (full version and abbreviated version of the questionnaire) did not have the same quality of evidence across measurement properties. Therefore, clinicians and researchers may consider the evidence for each measure since all measures from the same suite of tools have variable quality of evidence. In addition to assessing quality, measure selection needs to consider age group, target populations, and environment to ensure that measures are fit for purpose.

None of the studies reported on responsiveness for any of the 12 measures included in this review. Three of the measures were designed as screening tools (EPYFEI, SP2, and SSP2). When selecting a measure as a pre-/post-test measure, clinicians and researchers ought to ensure that the measure is designed as an outcome measure and not as a screening tool; there is no evidence, in terms of these studies, on responsiveness on any of the other nine measures.

It is interesting to note that half of the measures[49,50,56,60–64] included in this review were published in the past 6 years (between 2017 and 2020). This indicates that there is a growing interest and need for the development of new sensory-based measures.

Limitations: evidence

Studies in this review did not state if participants had commenced or received any previous sensory-based intervention or if study participants were stable at the time of recruitment or test–retest administration of questionnaires. Parents/caregivers whose children attended sensory-based interventions may have a more sophisticated understanding of sensory processing issues and heightened sensitivity to the behaviours associated with sensory input. Also, their children's behaviour may change due to therapeutic interventions.[66] This raises potential bias in reporting study results. Seven measures[47,50,54,55,57,58,64] were developed before 2018. However, modern psychometric measurement development has evolved since, such as the development of COSMIN standards for measurement development in 2018.[36–39] Since COSMIN emphasizes the need for adequate content validity of a measure, among other psychometric standards, these measures no longer meet the current standard for measure development.[36–39] Thus, highlighting the importance of ongoing evaluation of existing measures against the continuously improving criteria for measure development is needed to ensure that measures meet current standards.

Only two measures (SPM and SPM-P) had studies reporting on all of the 10 measurement properties. Most measures ($n = 8$) had only a single publication reporting on psychometric properties.

Limitations: review process

When developers elect to partner with a publishing company as part of test development, it limits the ability to publish in peer-reviewed journals. This creates a challenge when conducting systematic reviews since measurement manuals were not identified in a literature search across databases. We overcame this challenge by searching all known publishers and distributors of assessment measures. This study identified six measures that were excluded from the review because there were no published measure development, content validity, or psychometric studies for these measures (Table S8). Although these measures may potentially be psychometrically sound, they could not be included or evaluated in this review. Therefore, these measures should be used with caution due to the lack of evidence pertaining to content validity and psychometric measurement properties.

Although the term 'responsiveness' was used in the search strategy, no studies on responsiveness were retrieved for any of the measures. Since the PSEQ was the only measure to have adequate content validity, we recommend that an additional systematic review be conducted specifically to identify all studies that have used this measure in intervention studies. Meta-analysis on the pre-/post-test data to determine the responsiveness of this measure should be conducted.

Conclusion

To assess treatment efficacy, validated, sensitive, and reliable proxy-reported sensory-based measures are necessary. This review provides a guide for clinicians and researchers to aid the selection of these sensory measures.

It is imperative that, as part of measurement development, content validity studies are included to ensure comprehensibility, comprehensiveness, and relevance. Of the 12 measures included in this review, only three (EPYFEI, PSEQ, and SP2) provided studies on content validity. Of these three measures, the PSEQ-H had moderate quality for content validity studies but also had high-to-moderate quality evidence for sufficient psychometric properties that were tested. Although the other measures varied in quality across the other measurement properties, these should be used at the discretion of the clinician since measures without content validity cannot be recommended for use.[36–38]

This review highlights the importance of consumer involvement in the development of measures. Clinicians and researchers should consider content validity and psychometric measurement properties to ensure measures are fit for purpose.

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DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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SUPPORTING INFORMATION

The following additional material may be found online:

Appendix S1: Prisma-P Protocol.

Figure S1: Flow diagram of our approach to using COSMIN for conducting a systematic review of measures and COSMIN taxonomy and definitions.

Figure S2: PRISMA 2020 flowchart including all databases, registers, and other sources.

Table S1: PRISMA 2020 checklist.

Table S2: Example of search strategy for Medline via Ovid.

Table S3: Search strategies for all 11 databases used in this systematic review.

Table S4: COSMIN criteria for good measurement properties.

Table S5: Definitions of the four different ratings for the quality of evidence.

Table S6: Description of all included studies evaluated in this systematic review.

Table S7: Details the characteristics of the 12 included measures.

Table S8: List of excluded sensory measures from this systematic review and rationale for exclusion.

Table S9: Quality of measure development results using the COSMIN Risk of Bias checklist.

Table S10: Quality of measurement properties per study based on COSMIN quality criteria.

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CHAPTER SIX

Parent-young person lived experience of sensory dysregulation in children with tic disorders: Qualitative Study

Sensory dysregulation experiences are pervasive in children with tic disorders (Chapter 3, Study 1). Anecdotally reports from patients through Study 1 (Chapter 3) and Study 2 (Chapter 4) identified that sensory dysregulation experiences impacting the child/ young person function are not captured on most sensory measures. Thus, the breadth and extent of dysfunction are not accurately recorded or addressed in clinical practice. This fourth study aims to provide an understanding of the breadth of sensory dysregulation experiences and the impact on participation in daily tasks through the lived experience of young people with tic disorders and their parents.

This chapter explains the findings from a qualitative study conducted with 16 families with young people with tic disorders and their parents. This was the first step in a three-step process to develop a new patient-report sensory-based measure for use with children with tic disorders. The qualitative interviews allowed consumers to voice their experiences of sensory dysregulation and the impact this has on their lives. The next phase of the study will focus on generating the relevant items for the new measure through co-design and collaboration with consumers.

As this study has been sent to a journal for publication, this chapter is presented in the same format as a published paper. An authorship statement has been provided. Thereafter the

sections: abstract, background, methods, results, and discussion will follow. A list of references will be found at the end of the chapter. All supplementary documents made reference to in the manuscript are placed in the appendices of this thesis.

Authorship Statement

The co-authors of the paper ‘‘Parent-young person lived experience of sensory dysregulation in children with tic disorders: Qualitative study’’ confirm that ***Nicolette Soler*** has made the following contributions:

- Conception and design of the research
- Collection and extraction of all data
- Analysis and interpretation of the findings
- Drafting and revising the manuscript and critical appraisal of content

As the primary supervisor for the candidature upon which this thesis is based, I can confirm that the above authorship attribution statement is correct.

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Parent-young person lived experience of sensory dysregulation in children with tic disorders: Qualitative Study

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Parent-young person lived experience of sensory dysregulation in children and young people with tic disorders

Abstract

Purpose: To understand the experience of sensory dysregulation on participation in daily tasks for young people with tic disorders and their parents.

Methods: Eighteen semi-structured interviews were conducted with 16 families with children (5-16 years) with tic disorders and sensory dysregulation. Interviews ranged from 45 to 120 minutes and were transcribed verbatim. Thematic analysis using inductive and open coding methods was implemented to analyse the data precisely, consistently and exhaustively through recording, systematising and transparency of analysis to ensure trustworthiness.

Results: The impact of sensory dysregulation on daily life may be understood through one higher-order theme and three subthemes. The higher-order theme: ‘‘sensory, emotions and tics; it’s a ticking time bomb’’ emerged, and the three subthemes were: 1) we sacrifice and adapt to get daily activities done in the home, 2) my child’s experience of the community environment hinders participation, and 3) these sensory preference impact our entire family.

Conclusion: These sensory dysregulation experiences impact the entire family's quality of life. Additionally, the breadth of sensory dysregulation experiences are not captured in available sensory measures. There is a need for a patient-reported sensory-based measure for children with tics to comprehensively assess sensory dysregulation experiences sensitive to this patient population.

Keywords: sensory dysregulation, tic disorders, qualitative study, lived experience, paediatric

Implications for Rehabilitation:

- First qualitative study investigating sensory dysregulation experiences in paediatric tic disorders from the child and family perspective.
- This qualitative study provides a rich description of the range of sensory experiences and the impact on daily function.
- Provides insight into the interplay between sensory and emotional dysregulation and tic expression.
- Highlights the importance of understanding the impact of sensory dysregulation on the entire family unit.

Introduction

Tic disorders, a neurodevelopmental disorder, are the most common movement disorder in childhood, affecting 1 in 100 children [1]. Tics are repetitive, stereotypical, rapid, non-rhythmic movements or vocalisations which negatively interrupt a child's daily participation [2, 3]. According to the Diagnostic and Statistical Manual of Mental Disorders 5th edition (DSM-V), when both vocal and motor tics have been experienced in a waxing and waning pattern for longer than one year, the condition is referred to as Tourette's syndrome (TS) [4]. Co-existing neurodevelopmental and neuropsychiatric conditions are present in 80-90% of children with tic disorders [1, 5]. These include obsessive-compulsive disorders (OCD) (30-40%), attention deficit hyperactivity disorders (ADHD) (54-60%) and autism spectrum disorders (ASD) (5-15%) [1, 3, 6].

As reported in people with other neurodevelopmental disorders [7, 8, 9, 10, 11, 12], children with tic disorders and comorbidities experience sensory dysregulation [6, 13, 14, 15, 16, 17, 18, 19, 20, 21, 22] when compared with typically developing children [23, 24, 25]. Convergent evidence from electrophysiological studies [26, 27], magnetoencephalography [28], and neuroimaging [29, 30] indicate people with tic disorders and TS experience sensorimotor abnormalities. Deficits in sensorimotor gating, resulting in problems filtering irrelevant sensory stimuli, have been reported in individuals with TS [13, 27]. As a result of these sensorimotor abnormalities experienced by people with tic disorders, tic disorders should be recognised as a "sensorimotor" phenomenon rather than being understood to be pure movement disorder [1, 6, 15, 19, 26].

Several terms are used to describe these observed behaviours to sensory stimuli in children with neurodevelopmental disorders, including ''sensory dysregulation'', ''sensory processing'', and

‘‘atypical sensory reactivity’’ [15, 26, 31, 32]. Here we use the term ‘'sensory dysregulation’’ [15, 33, 34].

Sensory dysregulation is associated with reduced enjoyment and participation in daily life, including school engagement [24, 35, 36, 37, 38] and increased parental stress [9, 35, 39, 40]. Accordingly, the assessment and management of sensory dysregulation is an accepted part of comprehensive care for children with neurodevelopmental disorders [41]. Therapeutic approaches are commonly used to address sensory dysregulation in children with neurodevelopmental disorders, with most of these strategies having been developed for children with ASD [32, 42, 43]. Validated, sensitive, reliable, and responsive clinician-, teacher-, patient-, and proxy-reported outcome measures to assess treatment efficiency are necessary for clinical use in sensory dysregulation [44].

Our recent systematic review of a proxy-reported sensory-based measure for children with neurodevelopmental disorders found only one measure, the Participation and Sensory Environment Question Home Scale (PSEQ-H) [45, 46], satisfied moderate content validity [47]. The PSEQ-H was the only proxy-reported sensory-based measure from 12 measures evaluated to have consulted with consumers as part of measurement development to ensure the measure was comprehensive, comprehensible and relevant [47]. Yet anecdotal reports from paediatric patients with tics and their parents described sensory dysregulation experiences not recorded by either the PSEQ-H or the other commonly used proxy-report sensory-based measures [19, 47]. These reported sensory dysregulation experiences predominately relate to auditory sensitivities such as “people-made” noise, including cutlery scraping on crockery, chewing of food, or the “scratching” sounds of clothing [19, 47]. A recent study with children with tic disorders reported similar observations relating to experiences of auditory sensitivity to the same sounds made by

other people [48]. These tic-specific sensitivities to sound, in particular, are not captured in current patient or proxy-reported sensory dysregulation outcome measures [19, 47].

Therefore, a qualitative study was undertaken to understand the lived experience of sensory dysregulation experiences and the impact these have on the daily lives of young people with tic disorders. This study aimed to understand the breadth of sensory dysregulation experiences and the effect on participation in daily tasks by exploring the lived experience of young people with tic disorders and their parents.

Method

The study gained ethical approval through the Human Research Ethics Committee (2020/ETH00132) and Clinical Governance Committee (2020/STE00307) at the Sydney Children's Hospital Network, Sydney, Australia.

Patient and public involvement

The study protocol, as well as the interview guide, was developed with consumer consultation. The principal investigator (NS) met with two adults diagnosed with TS and sensory dysregulation to understand their lived sensory experiences, and the impacts of the sensory experiences on their daily life. From the knowledge gained through consulting with consumers, clinical experience with working with children with tic disorders and literature review, the study protocol and interview questions were developed [19, 49].

Research design

Using thematic analysis, the researchers identified, analysed and reported themes within the data obtained from semi-structured interviews utilising pre-determined questions [50]. Key features

from the extensive data sets were analysed and summarized using thematic analysis, permitting the perspectives of each study participant to be examined [50, 51]. Although an advantage to thematic analysis is the flexibility of the approach, the researchers conducted the study in a precise, consistent and exhaustive manner through recording, systematising and transparency of analysis to ensure the study's trustworthiness and the reported findings [51].

The pre-formulated interview questions explored participants' responses to their sensory dysregulation experiences and the impact on their daily lives and function (Supplementary documents 1 & 2). The wording and sequence of questions were left open-ended to support broader discussion and were guided by participant responses. In addition, study participants were provided with two prompt pages that used pictures and simple words to support discussions about lived experiences of sensory dysregulation and the impact on the person's activities of daily living (Supplementary documents 3 & 4). The semi-structured interviews were anticipated to be 30 - 60 minutes in duration. A demographic questionnaire was completed on the day of the interview.

Recruiting of study participants ceased once the research team agreed saturation of the data had been achieved. The data saturation point was understood to be once the same recurrent themes were identified through subsequent interviews, and no new themes were identified by additional participants [52].

Sampling and recruitment

Recruitment of study participants occurred through the Tic Clinic at a tertiary-level Children's Hospital in Sydney, Australia. The study recruited either i) parent(s) who had a child / young person with a confirmed tic disorder or ii) young people with a tic disorder and their parent(s). It

was at the family's discretion if the young person with a tic disorder would engage in the interviews.

The inclusion criteria for young people with tics to be eligible to engage in the study include: i) be between the ages of ten and sixteen years, ii) have a confirmed diagnosis of a tic disorder by a paediatric neurologist, iii) have known or suspected sensory dysregulation and iv) if prescribed pharmacotherapy, be on a stable medication regime for a minimum of six weeks prior to the time of recruitment. Study participants were not excluded if they had confirmed comorbidities in addition to a tic disorder diagnosis. Furthermore, the young person needed to vi) have conversational verbal skills to engage in the semi-structured interviews. Informed written consent from the young person and their legal guardian was obtained.

For parents or carers to participate in the study, they needed to be the carer of i) a child or young person between the ages of five and sixteen years with a medically confirmed diagnosis of a tic disorder. Eligible participants were sent information about the study and invited to participate via mail. After receiving a signed consent form, a convenient date for the family was set for the interviews.

Data collection

Due to the Covid-19 pandemic at the time of this study, all interviews were conducted via telehealth using Zoom software© [53]. All interviews were recorded with written consent from study participants and transcribed verbatim. All interview transcripts were uploaded and coded using NVIVO 12 software [54]. All study participants were provided with pseudonyms to protect their privacy and identity.

Data analysis

The descriptive qualitative data was analysed to determine the common themes concerning the participants' experience of sensory dysregulation and their impact on their daily function. The researcher used an inductive analysis approach as this was the best design for the question asked [50]. The themes were also identified within the coded data transcripts and were data-driven [50].

The researchers ensured the study's trustworthiness by being credible, transferable, dependable and confirmable [55] by implementing the six phases of thematic analysis, an iterative and reflective process, to analyse the data [50, 51, 56, 57]. These phases were: (1) familiarising yourself with the data, (2) generating initial codes, (3) searching for themes, (4) reviewing the themes, (5) defining and naming the themes, and (6) producing the report (figure 1) [51]. For a detailed description of the processes followed for each of these six phases, refer to Supplementary document 5.

To ensure the validation and trustworthiness of the findings, the principal investigator met with another occupational therapist with experience in qualitative research to review the themes and peer debriefing [51]. Study participants were asked to answer ten questions as part of the member-checking process to ensure that the themes reflected the study participants' experiences and echoed their voices (Supplementary documents 6 & 7). The questions ensured the findings relating to the themes were relevant, comprehensible and comprehensive and reflected the study participants' words and lived experiences [58, 59].

Three study participants provided feedback relating to member checking. Gaining knowledge that other children with tic disorders reported similar sensory dysregulation experiences as their child or young person highlighted for these study participants that they were not isolated in their

experiences. Through member checking, there was consensus on the comprehensiveness and relevance of all the themes. Regarding comprehensibility, members agreed with the wording used to describe all the themes except for the wording of the second theme by one study participant. This study participant, Karen (Interview PC011), explained that for them, it was Keri's perception or experience of the community environment that hindered her participation, not the environment itself. Following this insight and knowledge, the wording of the second theme was corrected to reflect this feedback.

Results: Participants

Of the 29 families invited to participate, 18 families chose to engage in this study. There was no reasoning provided to researchers as to why 11 families did not choose to be involved in the study. Of the 18 families who participated, two families were excluded as additional medical information at the interview resulted in the study participants being ineligible. Thus 16 families were included in the study. Two of these 16 families were interviewed twice, as a separate parent interview, and then a parent and young person interview was conducted at the families' request. Therefore, data was collected through interactive means as the principal researcher conducted all 18 semi-structured interviews with the 16 families engaged in the study. Ten interviews were conducted with the young person with tics in the presence of a parent(s), and eight interviews were conducted with parents of children/young people with tic disorders without the child/young person present.

The interviews ranged between forty-five minutes to two hours in duration. The mean duration of the interviews was 72 min. The demographic details of study participants are provided in Table 1 and Table 2. All names are pseudonyms to maintain confidentiality.

Results: Themes

One higher-order theme emerged: ‘‘sensory, emotions and tics; it’s a ticking time bomb’’ was established, and three subthemes were identified. These subthemes were: i) ‘‘we sacrifice and adapt to get daily activities done in the home’’; ii) ‘‘my child’s experiences of the community environments hinder participation’’; and iii) ‘‘these sensory preferences impact our entire family’’ (figure 2) (table 3).

Main theme: Sensory, emotions and tics, it’s a ticking time bomb.

The families have described an interplay between sensory and emotional dysregulation that exacerbates the child /young persons’ tics, resulting in reduced participation or engagement in tasks, reduced quality of life, and affecting the entire family unit. This interplay was described by Gretel, mother to Grace (Interview P007):

> *‘‘So, she's* [daughter Grace] *averse to heat. She hates the heat, and her tics go, um, like a hundred times worse in the summer. So does the rage. I think when she can't move that she gets more anxious, and then she'll tic more… Yeah. I think that's a big thing… Um. I honestly feel that if the sensory triggers weren't there, I think if they weren't so pronounced the tou, the Tourette's wouldn't be so bad. So, we see a huge increase in tics when she has to put clothes on. The whole neighbourhood hears us say F’ing C. Um.’’*

Keri, a young person with tics (Interview PC011), explained her experience:

> **‘‘***Breathing.* [the sound of other people breathing] *I think is definitely a trigger. It makes me very uncomfortable, and then it like goes and turns into a tic.’’*

Theme 1: We Sacrifice and adapt to get daily activities done in the home

Families live their life predictably and have become adept at identifying sensory triggers for their children. Parents and siblings avoid sensory stimuli, environments, activities, or events that may cause the child emotional distress resulting from sensory dysregulation. Parents also talked about adapting, accommodating and making sacrifices to ensure their child felt safe and comfortable to reduce sensory overload, emotional meltdowns, reduce tics and stress.

> Felicity, Flinn's mother (Interview P006): *''We learn to function. We're every day making the different meals and cutting off tags. It's just as soon as I buy his clothes now I know to have tags already off. And his dinner is always different to ours, as it has been for the last couple of years. So that's just part of our routine now. Very Stressful.''*

Within the home environment, the different tasks were broken down into (1) dressing, (2) mealtimes, (3) hygiene and grooming and (4) sleep which were problematic due to various sensory stimuli. It was also identified that participation in activities was affected by temperature, such as the ability to do homework. Each of these tasks will be explored in detail, and additional quotes to support the findings are provided in Table 3.

1.1 Dressing is a big challenge

Many aspects of clothing are known to be problematic from a sensory perspective, such as the texture or feel of the fabric, the clothing tags, seams in socks, wearing shoes and fabric stitching. These same challenges were reported by the majority (n=15) of the 16 families interviewed. Parents assisted their children in wearing clothing by cutting out the tags or purchasing clothing with the tag details screen printed on the fabric instead. Parents mentioned purchasing clothing items much too large for their child, which overcame the issue of the clothing feeling restrictive. Parents explained they had adapted the school uniform. In some cases, the children gained school

permission to wear the sports uniform over the formal school uniform, as this was more tolerable. Many children preferred to be naked than wear clothes.

> *Erin stated: (Interview P005): ‘‘Um, but we've just learned to adapt to it, and we just buy. I only buy now what I know she'll like, or I'll let her pick it. Yeah, undies are a pain. Always have been. So we cut the tags off the undies, and from Best and Less, we buy these seam-free ones where the actual tag is like a stamped onto it, as opposed to having a, an actual tag.’’*

These sensory challenges to clothing are well documented in the literature and rated on the current sensory assessment measures [60, 61, 62]. The findings from this study indicate that the implications of issues to dressing are broader than just problems relating to the feel of the clothing or fabric. Brian explained the impacts of his son Brett not being able to wear clothing (Interview PC002):

> *‘‘Use to make us late all the time because, you know, he just wouldn't find something that was comfortable, and it'd be an ordeal to get dressed. Yeah. Well, I took him to school in his undies once. It was that bad. [Laugh] Like, he just would not get dressed.’’*

To tolerate the continuous feeling of wearing clothing, families have adapted to having their child dress and undress multiple times throughout the day. Their child can only tolerate clothing for a limited time, such as when in public or essential. Nine-year-old Derek (Interview PC004) explains his routine of putting on clothing when leaving the house, but the moment he has the opportunity, such as in the car or at home, he takes off his clothes again.

‘‘*When my Nan comes over to pick us up, um, … I wait for the last second to go to the car and then put them* [clothes] *on. And then when I'm in the car, I take them off and then put them on* [to get out of the car] *and then at my Nan's house, I then take them off when I'm inside.’’*

For nine-year-old Grace, being naked in public is preferred over the sensation of clothing on her body, as described by her mother, Gretel (Interview P007). Due to the difficulty tolerating the feeling of clothing on her skin, her preference for being naked makes it impossible for her mother to have people come to the home to visit.

‘‘*Even if that means being naked in front of a crowd of people. Um, that's, that's a big thing. Cause after we leave the beach, I can't go and get milk or anything. We have to come straight home with a naked Grace in the backseat. Um, and what it means is that we're restricted to the home, and she doesn't wear clothes. So, people can't come in.’’*

The inability to tolerate the feeling of clothing outweighs the need to wear appropriate clothing. During the interviews, children explained that they would rather be cold than wear clothing. It was not that they did not feel the cold, but rather the intolerance of the fabric surpassed the need to be warm in cold weather. Annie, mother to 9-year-old Andrew (Interview P001), explained:

‘‘*He [Reference to son Andrew] really doesn’t like wearing clothes. He likes to walk around in his underwear, and he would do that year-round if we didn’t insist that he put something more on in the colder weather. Um, and in the colder weather even, he will only wear short sleeves* [Pause], *including to school. He wears the summer uniform year-round.* [Laughing] *He will have a shower or bath*

and then put [his] *underpants back on until he has to go out or somebody comes to the door. There'll be a mad rush.''*

When purchasing clothing, it was identified that money was wasted on clothing items children would not wear. The solution was only to buy clothing items chosen by the children. It has to be appreciated that taking children with sensory sensitivities to shopping centres to find clothing items of their preference comes with its challenges, which will be addressed in theme 2.

Brain (Interview PC002) explained: ''*So, this is the big one. You can't, can't buy clothes for him and say, here you go, you have to take him to the shop, and he has to try it on. He's got to feel the material. Then he will decide to buy it or if he wants it, and then we'll buy it. But if he turns around and says, I don't like the feel of it, even though it fits right, there's no point buying it because he won't wear it.''*

Although parents make financial and time sacrifices to ensure they can provide clothing their children can tolerate, it is evident that parents still feel shame at how their children are presented in public. The feeling of guilt is attributed to the concern that others may think they are not adequately providing for their children when they are not appropriately clothed in public. Although, in reality, their children are wearing the clothing they can tolerate, even if this means wearing old, worn-out clothes that no longer fit, clothing not suited to the appropriate weather or occasion, or scantly clothed with possibly no shoes, socks and so forth.

Gretel (Interview P007) stated: ''*So on a carer's pension when you buy clothes for your kid, but then when they won't, you've got to spend more money. And not only that, while you're waiting to find more money to spend and try and find clothes that they will actually wear, that's a really tough thing. You can't go anywhere... And that's really, really hard because I sacrifice a lot to give her*

really comfortable shoes. It's winter. And I just said, okay, my child is out at respite care with no shoes and no jumper. And everyone probably thinks I'm horrible and her t-shirts are disgusting.''

1.2 Mealtimes are very stressful

Mealtimes as a family is a very stressful occupation, resulting in family members eating separately to accommodate their child's sensory preferences. Families reported feelings of loss and being ''unconventional and different to other families,'' resulting in needing ''a different parenting approach to other families''. This is reflected in the quotes by the following families:

Maree (Interview P013) reflected: *''We have now gotten to the point where, and I know it's probably a really bad setting, but none of us sort of really eat together anymore because it's just too, too stressful.*

Food's texture, taste, and smell are known sensory issues relating to food sensitivities and often result in families having to cook separate meals. Two families engaged a dietician to assist with their concerns relating to their child's nutritional intake. A broader sensory issue relating to food was identified through these interviews. Families mentioned having to make accommodations around their child's intolerance to seeds in their food. Claire (Interview PC003), a young person:

''There's a lot of foods with seeds in them. I don't like food with seeds in them.''

In addition to these sensory challenges around meal times and food, families identified that the sounds relating to eating were problematic, resulting in stressful mealtimes and eating together as a family impossible. These include i) the sound of other people chewing food or swallowing ii) slurping a drink, or iii) the sounds of cutlery scraping on plates.

Keri (young person, Interview PC011) explained: ''*Anyone chewing sounds. Ah, sometimes it stops us from having family meals.''*

Sensitivities relating to food cause families to be unable to eat outside of the home environment, such as at restaurants or parties, due to the sensory input from the environment and the lack of acceptable food options for the child.

> Felicity, mother to Flinn (Interview P006), explained: ‘‘*Yes, so we've given up on going out to family restaurants. If me and my partner do want to go to a restaurant, it's just us two now because taking Flinn is just, but taking Flinn off to a restaurant, it's just a struggle.’’*

1.3 Hygiene and grooming tasks are overwhelming

Families reported that the morning and evening self-care and grooming activities cause their children distress and are emotionally overwhelming due to the sensory input experienced. These activities included but were not limited to bathing, washing hair, brushing teeth, and cutting their nails and toenails.

> Karen, mother to Keri (Interview PC011): *‘‘Actually, Keri finds just the whole package really stressful. And overwhelming, the whole the getting ready, all the steps, um, needed um, so as a whole it is quite stressful for Keri and feeling stressed and agitated and oh I got to get there on time, that is a big thing.’’*

Due to the distress and time these self-care tasks take to complete, families are frequently late for work and other activities. Many parents explained that they support their children in completing daily routine tasks by providing particular products to try and reduce the sensory issues their children experience.

For some families, challenges around self-care activities relate not just to the child partaking in the task but the child’s sensory experience of family members participating in self-care

activities. Inga's mother, Irma (P009), expressed the difficulty she and her husband, Ian, have with using deodorant, as Inga struggles with the smell of them applying deodorant. The lengths the family goes to accommodate Inga's needs were outlined.

> *''So even Ian's* [father], *Ian's deodorant. He can only put it on outside.* [Laughing] *Because when he puts it on inside, that's all she* [Inga] *can smell for half the day, and she complains about it the whole day. So* [Laughing]… *Even mine smells. He's tried a fair few different kinds, so it's not like he hasn't tried different kinds, she just doesn't like it, and I use a natural one.''*

Through these interviews, it was highlighted that for young girls who have reached puberty, the feeling of menstrual products is a problem, to the extent of impeding leaving the house or attending school.

> Karen (Interview P011): *''But now she is actually,* [she] *just got her period in the past couple of months, and so that is really, really challenging, she* [Keri] *can't stand the feeling of that. Yeah, I have tried so many things, and we find we have gotten onto a couple of products, but yeah, she's like so happy that it's lockdown* [reference to COVID-19 lockdown] *because she has got her period again.''*

1.4 It all has to be right to sleep

Several aspects concerning the sensory environment have to be controlled by parents to enable their children to feel comfortable and assist them in sleeping due to their child's sensory needs and sensory dysregulation. These include the feeling of the bedding, ensuring the fabric does not make a rubbing sound (such as when the material of the sheets rubs together), and the type of clothing or lack of clothing the child needs to wear.

Gretel, mother to Grace (Interview P007): *''Yes. Um, she didn't want me putting flannelette sheets on in winter because she likes the sheer cotton feeling. Um, but because she sleeps naked and only with one doona and I had to buy that doona in a hundred percent cotton and a certain type. And she makes me shake the, I have to shake the doona and make sure the quilt is distributed evenly every night, before bed. Mmm. And yeah, the sheets, she doesn't, she doesn't like me changing them very much. If the sheets change, everything is a problem. Anything that touches her skin is a problem. She doesn't have a top sheet because of that, so she just has a bottom sheet, and she has like, um, a light blanket and then a heavier blanket.''*

Parents aid their children to sleep by providing a sensory-supportive environment. This includes offering soothing sounds such as the sound of rain, ensuring the room temperature is comfortable, and the weight from the bedding is the right feel.

Karen, mother to Keri (Interview P001)**:** *''She just gets too hot, all the time, like she is always hot and, um, can't cope with it, it just feels so stuffy and um, just she doesn't sleep with very much on at all because she is just feeling hot all of the time. And um, and has to have a fan blowing on her all summer and often ice packs. Um yeah. Our bedroom is fairly close to Keri's bedroom, and she, even though she actually sleeps to a rain soundtrack to block any noise, and I am sure that she cannot hear Ken* [reference to Keri's father] *breathing through that, but she feels that she can, and so then she gets agitates and can't sleep, and so poor Ken has been on the couch for a few months now.''*

1.5 The heat affects my child's ability to engage in tasks.

The environmental temperature was not only identified to cause the child with sensory dysregulation discomfort but also impacted the ability of the child to participate in tasks and exacerbated their tics. One parent reported scheduling their child's tasks during the day to accommodate the weather, thus ensuring that the tasks may be completed in the cooler parts of the day. Gretel, Grace's mother (Interview P007), explained the observations in Grace's functioning when it is a hot day:

> *''Um, when it's hot, yes. It changes how our things are done. Um, she's* [Grace] *home-schooled. So. We have to try and do her schoolwork before she overheats and then just let it go for the afternoon. Um, everybody walks on eggshells. It's basically how can we manage not to piss off Grace but still get her to learn things like basic chores, some schoolwork,* [and] *attend appointments. It's, it's hard.''*

Theme 2: My child's experience of the community environment hinders their participation

2.1 Going shopping causes meltdowns.

Crowded places that are noisy and loud and embodies with multi-sensory experiences are challenging for children with sensory dysregulation. Such locations include shopping centres, parties, and social events. Due to children's emotional distress at being in these environments, parents explained that they frequently make accommodations by shopping online or avoiding events altogether. Not all shopping can be conducted online, especially when buying clothing for their child, as they need to feel the fabric to ensure that it meets their tactile needs, or else the clothing will not be worn. Gretel, mother to 9-year-old Grace, relayed their experiences (Interview P007):

> ''*And then the shopping and stuff too. That's off the wall,* [laughs], *you know. Um, yeah, that's hard. We have to go shopping sometimes, I can't do everything online, but the sensory stuff with that, it's not sensory seeking. It's more avoidance. So, she*

is constantly overwhelmed by loud noises as well. She's very loud herself. Um, and just being in the proximity of people. But when we went to our tiny little shop, she couldn't, she went in once and came out, and she just beat the crap out of me.''

2.2 We can't use public toilets.

Community outings need to be planned around their child's toileting needs, as for many families, their children are unable to access public toilets. Public toilets pose a problem due to the sounds made from the toilet flushing, the loud noise from the hand dryers and the smells.

Felicity (Mother P006): ''*He* [Flinn] *refuses to use public toilets… He just says they smell bad.''*

Penny, mother to Phoebe (Interview PC016): ''*I think noise has been a big thing for Phoebe from the beginning. She would block her ears when you flush the toilet and run away so she can try, so she can try, and block both ears whilst she presses the button.''* Phoebe (Young Person Interview PC016): *''Because it's always very loud. Um, like, I can't really be that far away when I flush the toilet. Although I do it a lot, it gives me a fright when I do it. Because of the noise level. Um, the yelling. Um, I feel a little bit freaked out about stuff. Yeah. I know, but it keeps scaring me.''*

2.3 Nope, no elevators, we work around it.

Escalators and lifts in community settings pose a problem to some children and young people because they fear heights and dislike the feeling of being on a moving surface. It is appreciated that parents accommodate their children's needs around using escalators and lifts in various ways, such as (1) using the stairs, (2) choosing different shopping centres without lifts or escalators, (3) parking on the same floor as the shop they want to access in a multistorey shopping centre or (4) supporting their children with strategies to manage to travel on the escalator or lift.

Maree (Interview P013) explains that she uses the escalator whilst her daughter Megan, uses the stairs to accommodate her daughter's dislike of escalators.

> *''Yes, definitely escalators. She would go to the stairs over an escalator if there was a choice next to each other. [Laughs] Because she does that at Sydney Central Station, she will go up the stairs. I'm just like, I'm going up the escalator.''*

2.4 We have to avoid activities with strong smells.

Smells that children find overwhelming or offensive result in children ceasing to partake in activities or be in particular environments. For instance, Keri (Interview PC011), a young person engaged in this study, refuses to attend appointments with her treating Ear, Nose and Throat Specialist due to the smell of his breath. Inga explains how accommodating her daughter Irma's sensitivities to smells in the environment have alternated and impacted their lives significantly (Interview P009).

> ''*If we outside and there's smoke or something like that, she gets quite upset with that, so smells do impact her, yes. So I will quite often avoid that area because I know how upset she gets, and then we trapped in a car, and it can be quite, um, hard for her, she gets, she, it doesn't work. Well, most of the time, we will move because I know that it just escalates. So, if we don't do something about it, it can just get worse, and she can get more uncomfortable and then, um, her anxiety can grow. I know it has definitely come up in our lives a lot. So, we have to alter our lives, yes.''*

2.5 I don't want to go to school; it's just all too much.

There are multiple facets to the school environment being recognised as a very challenging place for children with sensory dysregulation. Some of these factors were classrooms and the

playground being identified as too loud, the tactile element to certain activities, such as art and craft or sensory play, and this being offensive to children sensitive to getting their hands dirty and extra-curricular activities such as school discos being overwhelming. Attending school assemblies is a difficult environment for many of our study participants from a sensory and tic perspective. These difficulties relate to the loud and echoing environment, the expectation that children need to remain seated and still, the close proximity to other people in their space and having to suppress tics or being concerned that peers will notice their tics. As a result, parents explained that getting their children to attend school was difficult. In two cases, parents home-schooled their children so they could provide an environment to support their child's learning and sensory preferences.

> Heather, Jesse's mother (Interview PC010): *''It used to be hard to get him to go to school.''*
>
> Annie, Andrew's mother, explained (Interview P001): *''School assemblies are difficult for him. Generally, I would say the classroom is his least favourite place, although he really loves this teacher. But even with all those support strategies in place, he still finds the classroom difficult.''*

Theme 3: These sensory problems impact our child and family

Even though it is the child or young person who experiences the sensory dysregulation, the interviews highlighted the functional impact and toll which affects every aspect of the entire family's function, daily activity and quality of life. Karen, mother to 13-year-old Keri (Interview PC011), provided insights into how their entire family's lives are impacted by Keri's challenges with the sensory input from her environment:

> ''*It's horrible. I look at photos of how we were two and a half years ago, and I can't believe it's the same family* [laugh]. [Sigh]. *Like, uh, it's just awful, we can't*

go camping, we can't do that anymore. You know going in cars are really difficult, it's just awful, and we can't have a family meal together anymore. ... it just got worse, and I think now that has become, um and over time, it's just become morphed into Ken's [father] *eating and chewing and breathing and my swallowing sometimes. [Laughs]. And if I accidentally click my nails when I am when I'm driving the car.* [Laughs]. *And Ken's sleeping on the couch. We can't have a meal. So, it was a bit confusing. I don't quite understand what that means, and it's like, oh my God, our family is breaking up. We've all become so conscious of it that* [Laughs] *we just try not to swallow. We try to breathe shallowly.* [Laughs] *It's just ridiculous.''*

3.1 Leisure activities are stressful rather than relaxing.

Families explained that the television or radio volume is frequently a trigger for a child with sensory sensitivities to loud noises. As a result, the volume will be turned down, the show may be switched off, or an argument may ensue between the child and other family members. What was planned as a relaxing time for the family to spend meaningful time together inevitably becomes a stressful and disruptive event.

Karen (Interview PC011): *''Every Friday night, we'd have a family movie night, and um, we just noticed every time she* [Keri] *would just get really agitated then start yelling at her brother, who is two years older than her.''*

Families also mentioned that they noticed their child's tics worsen when watching television. As previously highlighted, a link between sensory stimulus and tic expression is apparent.

Penny (Interview PC016): **‘‘***So, I don’t know whether it is the noise or the flash of the TV, I don’t know what it is. But, ever since we have noticed this, uh, since the age of seven, well, when we first started with you guys as well, that was, um, definitely the biggest thing we noticed was in front of the TV, the tics got ten times worse. So, um yeah, whether it’s the noise or the lights, I am not sure. So. Yeah.’’*

Erin (Interview P005*): ‘‘It's so strange, the neck tic* [reference to daughter Emma’s tics] *is just once every, so often at night time and when she's tired and when she's home or watching TV, and she's quite relaxed, that's when you'll get seven or eight in a row.’’*

3.2 Car trips are challenging

Travelling by car or public transport is challenging due to factors such as the sounds from other passengers breathing, the car radio, the proximity of passengers to the child / young person, and the feel of the seatbelt or car seats being offensive. The inability to travel in a car or use public transport impacts not only functional activities such as attending school, medical appointments or community activities but also being able to go on holiday. Families try to work around the sensory issues their children experience in these settings by using headphones, not having the radio or music playing, avoiding long car trips, and using private vehicles instead of public transport. Karen, mother to Keri (Interview P011), highlighted that the family are unable to go on holiday as their daughter is too distressed by travelling in a car or on public transport due to the sounds made by other people around her, including people breathing.

‘‘You know going in cars are really difficult, … um we can’t go on family holidays uh she has to um, car trips are really difficult um she has to have headphones on…

Um the car would be very challenging [due to the sound of people breathing]. Um, but uh, she found the travel on public transport overwhelming and couldn't cope with the crowded carriages.''

Brian, Brett's father (Interview PC002), explained: *''But I think constant sound also in the car. Like, if we crank up the music, the other son and me love it, whereas Brett's, like, blocked ears, too loud. Got to turn it down.''*

3.3 We just stay at home; events are a no-go.

All families reported that their children with sensory dysregulation felt most comfortable and preferred to stay home. This resulted in families not being able to engage in community activities the same way other families would.

Maree, mother to Megan (Interview P013), explained: ''*I suppose it has just been our norm, um, like because nowadays she just, yeah, won't really go anywhere. She just wants to be at home.''*

Families experienced social isolation as not only could they not engage in community events or activities, but hosting friends and family at their homes was not even possible.

Karen (Interview P011): *''Loud noises, um, are an issue. She* [Keri] *has always been really like* [that] *all of her life actually, just very worried about all fireworks… We can't go out to see friends as much anymore or have friends over.''*

When going to a music concert, Brian *(Interview PC002)* mentioned that his son Brett became upset as the environment was too loud:

''Concerts, yeah. He doesn't like loud music, let's say we went to the U2 concert last year and it was a bit of a struggle with him. He got upset. I think it was a bit

too much, we. We weren't right in the middle of it. We're sort of in the stadium towards the back.''

A discussion between Harry and his father, Hamilton, during interview PC008, provided insight into how parents are aware of and protect their children from the triggering sensory stimuli in the community environment:

Harry (Young Person): ''*When it's too loud. It's a small space, and the sound waves echo through… Well, I don't avoid.''* Hamilton (Father): ''*Yeah, well, you sort of do. I've got to protect you from them. You run away. You put your hands over your ears.''*

3.3 The rage & outbursts, we're walking on eggshells.

Emotional outbursts by children and young people were described profusely by parents when there was either a sensory trigger or a build-up of sensory input experienced. Derek and his father, David (Interview PC004), explained that as the day progressed, Derek became overwhelmed by the various sensory experiences involved in his daily routine, causing emotional dysregulation and resulting in family arguments.

David*: ''When you have a bad day, is it like all different things that are, like one thing might annoy you and then another one annoys you more, and it sort of builds up, and then everything explodes over the top.* Derek*: ''Yeah. Cause, like, the other day, um, my clothes, I didn't like my clothes, and then, I had an argument with my dad, mom, brother, and nan.''*

Families voiced the impact these emotional outbursts experienced by their children have on participation in activities, quality of life, family togetherness, and relationships. Gretel, mother

to Grace (Interview P007), details the impact Grace's emotional outbursts have on their family and the lengths she goes to support Grace to stay calm*:*

> *''it's hard because I restructure everything. Everything to make things less stressful for her because if she's not stressed, I'm not stressed. But when Grace is stressed, the iPad gets thrown at me. Holes get put in walls. The baby gets hurt. I get hurt. Then she thinks she's the worst person in the world and hates herself. And it's just a cycle, and it's horrible. So, yeah, I do very much change everything.''*

Besides the sensory triggers, parents described their sensory-sensitive children to anger very quickly, and emotional dysregulation was very common. Multiple parents reported that their children became physically violent when angered. Brett, a young person (Interview PC002), described his challenges with emotional dysregulation:

> Brett: *''Yes. Yes, I get angry easily, yeah. Yeah. Yes. Controlling my emotions is very hard. And it does get a bit tricky doing that.*

Some families reported that even though their child's sensory preferences caused sensory dysregulation, impacting the quality of life and function, it was actually the behaviours resulting from emotional dysregulation that was most challenging.

> *Gretel (Interview P007): ''Um, everybody walks on eggshells. It's basically how can we manage not to piss off Grace but still get her to learn things like basic chores, some schoolwork,* [and] *attend appointments? It's, it's hard. It's like she has this internal engine that is just going, going, going, going, and it only has one gear, well, maybe one or two gears, but not enough to slow down by yourself sort of thing. Once she, once she revs up, she can't get back down, and that's actually,*

that's, that's a major thing. So, when you try and take that away (IPad), people get injured. The anger and the rage is so big and horrible that I don't get to see the other sides of her as much as I'd like to. That's really hard.''

Laura, mother to Liam (Interview PC012), explains the impact Liam's anger has on the family unit and on his bother too*:*

''He just gets really angry. I mean really angry. Like people wouldn't believe me how angry he can get, he gets really angry… its starts a lot of the time it starts with frustration. And it could last maybe, you know, half an hour… That we can get into these spirals where we, you know, we've just go and lie down or do something until we can bring ourselves back to where you can actually function. So that, that's probably has a massive impact on our, on our lives. Well, the family unit. I mean, he's got a younger brother who tends to, you know, bear the brunt of it. It's just not a nice environment. Like when we are all yelling at one another, and um, you know where you worry that you know his head is going to come off because he's so angry.''

Discussion

The impact of sensory dysregulation on function, participation and quality of life resulting from the child experiencing sensory dysregulation affects the entire family, not just the child or young person. In many cases, the child's sensory needs are accommodated by the parents providing an environment that facilitates the child or young person's sensory needs and preferences, ensuring harmony in the home. We completed 18 interviews, and overwhelmingly, all families reported the impact of sensory dysregulation on all aspects of their child and young person's life and the quality of lives of the entire family.

Parents have learnt to understand their child's sensory needs. They adapt the environment and the tasks so their children can successfully complete tasks. Not all environments can be adapted, so some families must make sacrifices impacting their quality of life to meet their children's or young persons' needs.

The impact on the family ranged from being unable to eat meals together to being unable to listen to the radio in the car or watch television without upsetting their child with auditory sensitivities. The noises made by family members, such as chewing and swallowing sounds, scraping of cutlery on plates, breathing noises and clicking of fingernails, cause disharmony in the family and impact the family togetherness. A case series stated that auditory stimuli caused emotional outbursts and increased tic expression due to misophonia (sensitivity to certain sounds causing an extreme emotional response) in children with tic disorders [48]. This case study by Robinson et al. (2018) reported similar auditory sensitivities to human-made sounds as was identified through the qualitative interviews in this study [48]. In addition, the effects of temperature on the child's ability to engage in tasks, the difficulty with tolerating the feeling of seatbelts and car seats, and the inability to travel in a car. Hence family holidays are impossible or very difficult, due to the difficulty travelling together.

Studies by DeGace (2004) and Fernansez-Andres et al. (2015) found that families with children with severe ASD may experience difficulty engaging in daily activities that hold positive meaning for them due to sensory needs and rigid routines [63, 64]. Children with ASD and auditory filtering difficulties were 47% more likely to underachieve academically in the absence of intellectual difficulties than neurotypically developing children [65]. Fine and gross motor difficulties correlated with sensitivities to visual, tactile and movement stimuli on the Short

Sensory Profile impacting motor performance in elementary school children with ASD[66]. Children with ADHD and sensory dysregulation were found to participate in less leisure activities than neurotypically developing children, and it was recommended that evaluation and intervention programs enhance the child's relationships with peers and the child's well-being [67, 68]. In children with high-function ASD, sensory dysregulation (in particular oral and touch sensory systems) was the strongest predictor of social impairment [69, 70]. Ritual behaviours may be developed as a coping mechanism in response to anxiety experienced by children suffering from sensory dysregulation [23, 71]. Intervention for sensory dysregulation focuses on supporting the child rather than facilitating the occupations and needs of the entire family unit [63]. As the family unit provides a valuable source of learning and development for a child with neurodevelopmental disorders, it is prudent that, as clinicians, we support the family to expand and enhance these opportunities for their children [63].

Parents voiced that addressing their child's emotional dysregulation in therapy was paramount, over and above the challenges of sensory dysregulation. This need to address emotional dysregulation over and above the sensory dysregulation experiences may be because families have learnt to be insightful regarding their children's sensory preferences and how to accommodate them. Study participants also mentioned an interplay between sensory dysregulation, emotional dysregulation and tic expression. Research on the prevalence of sensory dysregulation and tics [15, 19] is emerging, and the effects of emotional dysregulation and tics, such as factors such as stress, anxiety, excitement and fatigue, exacerbate tics is well documented [72]. Yet the interplay between sensory and emotional dysregulation and tics is to be further understood.

Therefore, as clinicians and researchers, it is valuable to understand and measure the breadth of sensory dysregulation experiences children with tic disorders experience to ensure a comprehensive assessment of the issues. This is to aid in determining the severity of the impact on function, assist with formulating treatment planning and measure treatment outcomes. The impact of sensory dysregulation experiences on the family unit as part of the assessment should be considered to ensure that therapy focuses not only on improving the function of the child, but the entire family unit. Quantifying the strategies used by families needs to be captured as these provide valuable data on how best to support children and provide an understanding of the implications these accommodations have on the quality of life for the entire family unit [63].

The interplay between sensory dysregulation, emotional dysregulation and tic expression ought to be considered. In a case series of twelve children with tic disorders, the researchers hypothesised that sensory sensitivity to sound (misophonia) caused abrupt emotional dysregulation in individuals with tic disorders and should be considered part of a comprehensive clinical assessment. [48]. Furthermore, children with tic disorders and comorbidities have been identified to experience sensory and emotional dysregulation [15, 73]. Tic expression is significantly related to stress and emotional dysregulation by various emotional states [74, 75, 76]. It is suggested that treatment focusing on the mediating role of emotion dysregulation may contribute to developing improved therapies for children with tic disorders [75]. Therefore, sensory and emotional dysregulation and tic severity should be considered through the assessment and treatment process to ensure a holistic and comprehensive understanding of the patient's needs and condition.

In addition, the researchers acknowledge that 80-90% of children with tics experience other neurodevelopmental or neuropsychiatric disorders [1, 3, 6]. All the study participants

experienced at least one other comorbidity in addition to a confirmed tic disorder (Level 1ASD: n=4, OCD: n=5, ADHD: n=7 and anxiety: 12). Even though all study participants (n=16) experienced sensory dysregulation, these experiences could be at least partly be influenced by another comorbidity such ASD (need for sameness, restricted eating), OCD (need for things to be same, just right), and anxiety (how sensory issues trigger emotional dysregulation) [4]. But this is a 'reality' of children with neurodevelopmental disorders, who rarely have a single disorder, but often have multiple comorbidities. All the study participants reported these sensory dysregulation experiences. Thus, they all were diagnosed with a tic disorder (n=16) and were not restricted to children with any specific comorbidity, i.e., ASD or OCD.

Limitations of the Study

Although inductive analysis was the predominant approach to data analysis, it is understood that researchers could not free themselves of their theoretical and epistemological commitments [50]. The researchers had prior theoretical knowledge and experience in paediatric tic disorders, having conducted previous quantitative research studies [19, 47, 49] and worked clinically in this field. To overcome this bias, the following steps were taken to address, which included researcher triangulation, peer debriefing, themes and subthemes being vetted by team members and member checking to ensure that the research themes are trustworthy and credible.

Future research:

As the current proxy-report sensory-base measures focus only on rating the sensory dysregulation of the child [47], there is a need for the development of a new measure that comprehensively assesses the breadth of sensory dysregulation experienced by children with tic disorders and comorbidities as identified through these lived experiences [59, 77, 78, 79]. In addition, the measure needs to allow for the impact of sensory dysregulation on the family

unit and an understanding of the interplay of sensory, emotional dysregulation and tic symptoms. This would not only aid in approved assessment and treatment planning but also allow for rating the effectiveness of treatment through the clinical utility of comprehensive, comprehendible and relevant sensory-based assessment measure for children with tic disorders and comorbidity.

Conclusion

Children with tics and their families experience impacts on their participation in daily tasks due to sensory dysregulation that often remains unmeasured and a resultant lack of treatment options due, in part, to the hidden nature and lack of awareness of these issues. Families have reported incredible resourcefulness in adapting to the challenges. Still, a holistic response is needed to manage the impact on function resulting from these sensory dysregulation experiences. A comprehensive evaluation of the effects of sensory dysregulation in children with tic disorders needs to consider a broader list of experiences than is currently being assessed in current practice. The assessment needs to provide insight and measurement of the accommodation and adaptions made by the family and the impact of sensory dysfunction on the entire family unit. A comprehensive, comprehensible, relevant sensory-based measure is required to effectively assess and treat children and young people with tic disorders.

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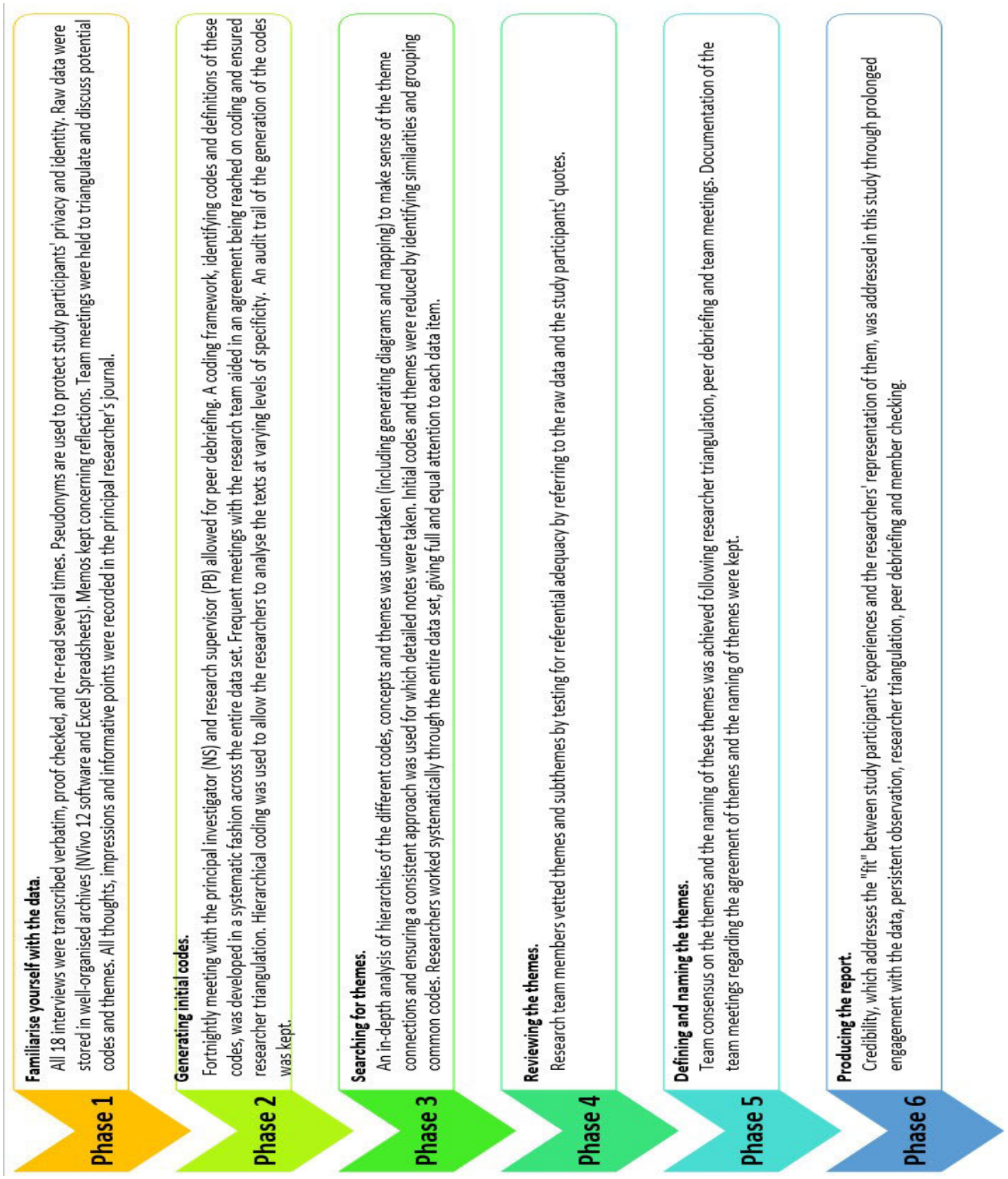

Figure 1: A diagram depicting the 6 Phases of thematic analysis used in the method of data analysis of this study and a description of the activities completed by the researchers for each phase [50, 51, 57]

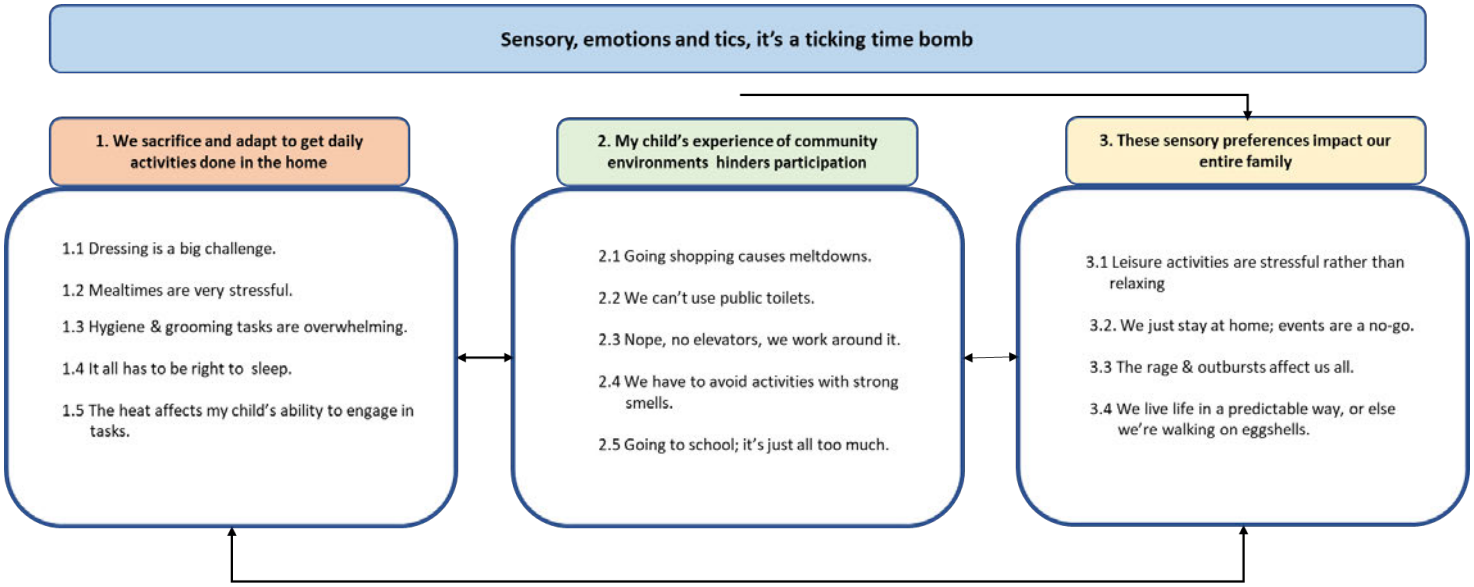

Figure 2: A diagram depicting the themes identified in this study. There is one higher-order theme: ''Sensory, emotions and tics, it's a ticking time bomb'' and three subthemes: 1) We sacrifice and adapt to get daily activities done in the home, 2) my child's experience of community environments hinder participation and 3) these sensory preferences impact our entire family.

Table 1: Demographics of study participants, including parents and children and young people engaged in the study.

Demographic details of parents interviewed		
Characteristics	All Parents interviewed (n=19)	Parents of young people interviewed (n=13)
Gender (Female, Male)	F=15, M=4	F=9, M=4
State:		
ACT / NSW/ VIC	3 / 12 / 1	3 / 6 /1
Level of income: *		
Most Financially advantaged	7	5
Financially advantaged	1	0
Between advantaged and disadvantaged	6	5
Financially disadvantaged	0	0
Most Financially disadvantaged	2	0
Marriage Status of families:		
Married	11	7
Defacto relationship other than biological parent	2	1
Single	2	2
Employment status of parents interviewed		
Paid employment	14	11
Carer to child	5	2

Demographic details of children and young people		
Characteristics	**All Children/ young people in study (n=16)**	**Young people Interviewed (n=10)**
Gender (Female, Male)	F=9, M=7	F=4, M=6
Age: Mean age (years & month)	11, 5	12, 2
Range (years & months)	7, 4 – 15, 11	9, 2 – 15, 11
Primary diagnosis of Tic disorder:		
Tourette's Syndrome	14	9
Chronic vocal tic disorder	1	1
Chronic motor tic disorder	1	0
Average age of onset	4 years, 4 months (Range: 2 years – 10 years)	4 years, 3 months (Range: 2 years- 10 years)
Secondary diagnosis: *	16	10
Allergies	1	0
Anxiety**	12	7
Attention Deficit Disorders:	7	5
Inattentive subtype	6	5
Hyperactive subtype	1	0
Autism spectrum disorder (Level 1)	4	1
Dyspraxia	2	2
Eating Disorder	1	0
Eczema/skin conditions	3	2
EDS	2	0
Emotional Disorder	16	10
Insulin resistance	1	1
Mood disorder	4	2
Obsessive-compulsive disorder	5	4
Oppositional defiance disorder	2	1
PANS	1	0
Sleep disorders	3	1
Speech difficulties***	5	1
Taking prescription medication	13	9
Specialist Medical Care:		
Cardiology	1	0
Neurology	16	10
Psychiatry	2	1

Characteristics	All Children/ young people in study (n=16)	Young people Interviewed (n=10)
Participating in therapy:	12	7
Dietician	2	0
Occupational Therapy	9	5
Physiotherapy	1	0
Psychology	11	6
Speech Therapy	5	1
Year of education: Mean & Range	Year 5 (3-10)	Year 6 (3-10)
Type of schooling:		
Mainstream without support	2	1
Mainstream with support****	12	9
Homeschooled	2	0
Handedness: Right / Left	15, 1	10, 0
Premonitory Urge experienced (PU)	12	6
No PU experienced	2	2
Unsure if PU experienced	2	2
Have siblings	16	10
Siblings have a diagnosed NDD*****	9	6

ACT (Australian Capital Territory), NSW (New South Wales) and Vic (Victoria). The level of income is based on the study participants' post-code in relation to the information provided by the Census of Population and Housing: Socio-Economic Indexes for Areas (SEIFA), Australia, 2016.

*As per Diagnostic and Statistical Manual 5 (DSM-5) [4] criteria for comorbidities.

** Anxiety disorders include all forms of anxiety, such as 92generalised anxiety, social anxiety, separation anxiety etc.

***Speech difficulties included stutter, articulation, pronunciation or social skills training

****School support includes the provision of additional time for classwork or exams, special accommodations such as writing exams in a different venue, ability to leave the classroom to tic, additional learning support officer involved, use of therapeutic strategies in the classroom etc.

***** NDD= Neurodevelopmental disorder.

Table 2: Details of each interview with each of the 16 study participants.

Interview	Parent(s) (Pseudonym)	Parent's Gender	Second Parent(s) (Pseudonym)	Second Parent's Gender	Child (Pseudonym)	Child's Gender	Child present	Interview duration (min)	Primary Diagnosis	Age of onset (years)	Medication
*P001	Annie	F					N	89	TS	5	N
PC001					Andrew	M	Y	76			
PC002	Brian	M			Brett	M	Y	62	TS	3	Y
PC003	Carmen	F			Claire	F	Y	58	TS	5	Y
PC004	Debbie	F	David	M	Derek	M	Y	71	Chronic motor tic	2	Y
P005	Erin	F			Emma	F	N	78	TS	4	Y
P006	Felicity	F			Flinn	M	N	46	TS	3	Y
P007	Gretel	F			Grace	F	N	114	TS	8	Y
PC08	Heather	F	Hamilton	M	Harry	M	Y	67	TS	5	Y
P009	Irma				Inge	F	N	52	TS	3	N
PC010	Heather	F	Hamilton	M	Jesse	M	Y	116	TS	3	Y
*P011	Karen	F					N	73	TS	10	Y
PC011					Keri	F	Y	70			
PC012	Laura	F			Liam	M	Y	56	TS	3	Y
P013	Maree	F			Megan	F	N	80	TS	3	Y
PC014	Naomi	F			Natalie	F	Y	66	TS	4	Y
P015	Olga	F			Olivia	F	N	47	TS	6	N
PC016	Penny	F			Phoebe	F	Y	67	TS	3	Y

The interview is coded as either PC or P. PC identifies that both the parent and the child were present in the interview, and a code of P identifies only the parent was present. * Two families were interviewed twice as the parents wanted to be Interviewed in the absence of their child (P001 & P011) and then again with their child present (PC001 & PC011). One family had two children with a tic disorder, and both children participated in the study (PC080 & PC010). All study participants identified as being Australian in relation to their ethnicity, except the families of PC080 & PC010, who identified as European. All children/ young people engaging in this study had a primary diagnosis of Tourette's syndrome (TS) except Derek, who was diagnosed with a chronic motor tic disorder.

CHAPTER SEVEN

Summary of Findings and Concluding Remarks

This final chapter provides an overview and summary of the knowledge gained from all four studies that comprise this research project and thesis. An overview of the research aims, and the method for each study is reiterated, followed by a discussion on major findings, and implications for clinical practice are highlighted. The limitations of the studies are reviewed, followed by a discussion on implications for future research directions and concluding statements.

79.1 Overview of the Research

As clinicians working with children with tic disorders, it was apparent that our patients were experiencing sensory dysregulation that impacted their function, which was not being assessed or addressed as part of standard care. Our patients reported struggling with wearing clothing due to the feel of the fabric, the seams or the clothing feeling restrictive or too tight. This impacted children's ability to wear school uniforms and even required some children to be home-schooled. Patients mentioned their children experiencing issues with sleeping due to the feeling of the bedding, and difficulty with mealtimes due to the texture of food and the sounds of people chewing around them. One patient even described being unable to travel in a car as he could not tolerate the feeling of the car seats and seat belts on his skin. This impacted his ability to attend school and medical appointments or engage in the community.

From our clinical patient observations and their reports relating to sensory dysregulation and the impact on function, and with limited literature to inform our practice (25), there was a need to better understand the prevalence of sensory dysregulation experiences in children with tic disorders. Additionally, there was a lack of understanding of how best to assess and treat children with tic disorders attending the Tic Clinic at the tertiary paediatric hospital, as a component was missing from the multidisciplinary care, with occupational therapy not included in the care team managing tics. The clinical practice guidelines relating to the assessment and treatment of children with tic disorders and comorbidities do not mention or consider sensory dysregulation in children with tic disorders (4, 55, 57). It is not standard practice for occupational therapists to work with children with tic disorders for tic management. In the few settings internationally where occupational therapists are employed in paediatric tic clinics, CBiT intervention is provided rather than sensory-based intervention to manage tics (25).

Furthermore, it was interesting to note that patients had identified for themselves and explained at their therapy appointments that certain sensory-based activities improved their tics. One child wore tight cycling gloves to reduce a tapping tic he did with his fingers. He mentioned that he liked the feeling of tightness on his hands. Chewing on a cold washer helped another child with a vocal tic.

Therefore, from our patient reports and clinical observations of children with tic disorders, it was evident that there was a need to understand the prevalence of sensory dysregulation in children with tic disorders. The prevalence of sensory dysregulation in children with tic disorders was poorly understood. Therefore, I lead a research study with 102 children with tic disorders and 61 healthy controls (Figure 7.1, Study 1) to scope the prevalence of the problem.

There was also a need to develop more global tic-related interventions (55, 60). The reports from our patients of self-identified sensory-motor strategies that effectively reduced their tics required further investigation as a form of a possible new multi-modal treatment approach to manage tics. This resulted in a pilot study (Figure 7.1, Study 2) of ten children to investigate if a sensory-motor-based treatment approach would effectively reduce tic intensity and frequency.

In addition to the study findings from these first two studies (Studies 1 and 2), it became apparent that study participants reported sensory dysregulation experiences that the researchers were unaware of. When reviewing the sensory dysregulation experiences rated on the two commonly used proxy-report sensory-based measures, the Child Sensory Profile 2 (CSP2) (Study 1 and 2) (155) and the Sensory Processing Measure (SPM) (Study 1) (26), many of our patients reported sensory dysregulation experiences that were not being rated or assessed. It

became evident that our patients' sensory dysregulation experiences were broader than the scope being assessed on these commonly used proxy-report sensory-based measures. Therefore, this highlighted a need to identify all the proxy-report sensory-based measures and determine which of these was the most valid, reliable and fit-for-purpose measure for use with children with tic disorders. Consequently, a systematic review of all proxy-report sensory-based measures used for children and adolescents with tic disorders and associated neurodevelopmental disorders was undertaken (Figure 7.1, Study 3). From the knowledge gained about the breadth of the sensory dysregulation experiences from patient reports and the findings from the systematic review, it was apparent that the current sensory-based measures may be inadequate to comprehensively assess the scope of the problem experienced by children with tic disorders in our clinic.

To evaluate if the commonly used proxy-report sensory-based measures are comprehensive and relevant for use in children with tic disorders, a qualitative study (Figure 7.1, Study 4) was conducted. Knowing if the current sensory measures were appropriate for children with tic disorders was imperative for clinical practice. Assessment measure findings guide treatment planning, goal setting, and intervention and are used (in many cases) as an outcome measure to ensure the effectiveness of treatment (156, 157). Thus an appropriate, relevant, comprehensive and comprehendible measurement tool must be used with patients (156, 157).

Hence, through conducting the four research studies mentioned above and presented in this thesis (Figure 7.1), we aimed to gain evidence to assist in understanding the prevalence of sensory dysregulation in children with tic disorders. In addition, the research aimed to aid in better understanding how to assess and treat our patients, who are children with tic disorders and comorbidities.

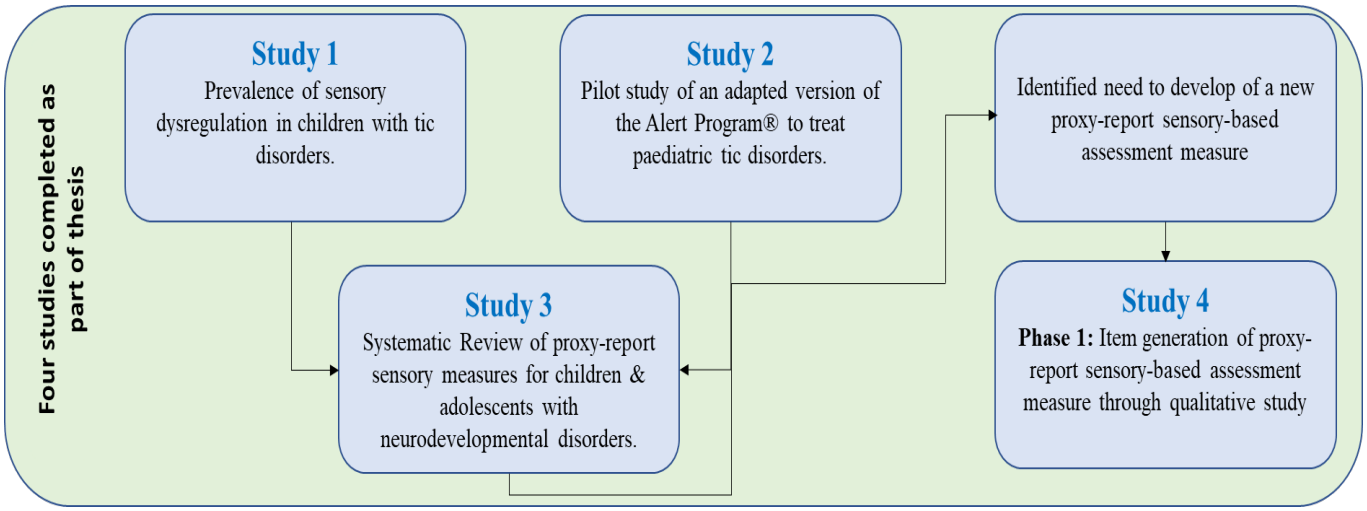

Figure 7.1 Flowchart of the four research studies conducted as part of this thesis. The diagram depicts the order in which the studies were conducted and how it emerged through the findings from Studies 1, 2 and 3 that there was a need to develop a new sensory-based measure (Study 4).

Four research aims were developed from the above-identified needs:

1. To measure the prevalence of sensory dysregulation in tic disorders and their clinical associations in a quaternary clinic sample (Study 1 and Chapter 3).
2. To pilot test the effectiveness of a sensory motor-based therapy to treat tic disorders in children (Study 2 and Chapter 4).
3. To determine the quality and utility of proxy-reported sensory-based measures for children and adolescents with neurodevelopmental disorders (Study 3 and Chapter 5).
4. To understand the breadth of sensory dysregulation experiences and functional impairment in daily life by interviewing young people with tic disorders and their parents (Study 4 and Chapter 6). This was the first step in developing a new sensory assessment measure.

79.2 Discussion of Main Findings

79.2.1 Children with Tic Disorders and Comorbidities Experience Significant Sensory- Dysregulation

The first study (Study 1, Chapter 3) (Figure 7.1.) aimed to define the prevalence of sensory dysregulation in children with tic disorders through a case-controlled cross-sectional study conducted between January 2017 and April 2018. Consecutive tic clinic patients (n=102) between the ages of five years to twelve years and eleven months (mean age nine years, five months) were recruited into the study and compared with sixty-one age-and-sex-matched neurotypically developing children. Sensory dysregulation, executive function, and quality of life data were obtained through the Short Sensory Profile-2 (SSP2), Child Sensory Profile-2 (CSP2), Sensory Processing Measure (SPM), Behaviour Rating Inventory of Executive Function-2 (BRIEF-2), Strength and Difficulties Questionnaire (SDQ) and Pediatric Quality of Life Inventory (PedsQL). For children with a tic disorder, a paediatric neurologist assessed tic severity using the Yale Global Tic Severity Scale (YGTSS).

There was a strong positive correlation between the scores of both proxy-report sensory-based measures, the SSP2 and SPM sensory assessments ($n = 162$; $rho = 0.842$; $P < 0.001$). When comparing sensory dysregulation (using SSP2 total raw scores), participants with tic disorders ($n = 101$) had elevated scores compared with healthy controls ($n = 61$; $P < 0.001$). There was a difference in sensory dysregulation scores on the SSP2 for participants with tics and comorbidities ($n = 87$) compared to those children with tics only ($n = 14$; $P < 0.001$). The presence of comorbidity was positively correlated with elevated sensory dysregulation, as shown for ASD, ADHD, OCD and any emotional disorder. In the total tic cohort compared to neurotypically developing controls, all SP2 sub-scores (i.e., seeking, avoiding, sensory, bystander, auditory, visual, touch, movement, body position and oral, conduct, social-

emotional, and attentional) were elevated (Chapter 3, Study 2, Table 2). As eighty-seven study participants with tic disorders had at least one comorbidity, and twenty-eight participants had three or more co-existing neurodevelopmental comorbidity, the association between sensory dysregulation and comorbidity needs to be appreciated. There was an increase in sensory dysregulation mean scores with an increase in the number of comorbidities (SSP2, n = 101; 5.4 units per comorbidity; 95% confidence interval [CI]: 4.4–6.4; P < 0.001).

In addition to identifying an increased prevalence of sensory dysregulation in children with tic disorders and comorbidities, an increased prevalence of executive function difficulties (BRIEF-2 Global Executive Composite [GEC]) in children with tics (n=101) compared to healthy controls (n = 61; P < 0.001) was apparent. In the tic group compared to the controls, all BRIEF-2 sub-scores were elevated, including the Behavior Rating Index (BRI), Emotional Regulation Index (ERI), and Cognitive Rating Index (CRI; Supporting Information Table S1; Supporting Information Fig. S2–S4).

It is interesting to note that children with tic disorders experienced significant differences in emotional dysregulation, which was supported by the anecdotal evidence through the 18 study participant interviews in study 4 (Chapter 6). So, study participants in study 4 reported that the emotional dysregulation symptoms were far more impairing than the sensory dysregulation symptoms.

79.2.2 A Multi-Modal Sensory-Motor-Based Treatment Approach Assists in the Treatment of Tic Disorders

There was a need to explore additional treatment approaches for children with tic disorders that complement existing treatment practices (55). The researchers tested a new treatment approach

to manage tics by pilot-testing a modified version of the Alert Program® (Chapter 4, Study 2), which uses sensory-motor strategies to assist children and adults with self-regulation and attention skills. The aim was to determine if a sensory-motor treatment approach, such as the Alert Program®, with children with tic disorders could reduce tic severity and frequency.

A sensory-based treatment approach was selected due to the ‘‘sensory phenomena'' often described by people with tics, including premonitory urge, 'just-right' perceptions or somatic hypersensitivity associated with tic disorders (25, 158). Clinically, several of our patients had identified sensory-motor strategies that reduced tic expression for themselves. However, this had not been prescribed by a clinician or reported in the literature. In addition, knowledge gained from our first study that children with tic and comorbidities have significant sensory dysregulation, a sensory-based treatment approach seemed appropriate.

Twelve participants diagnosed with tic disorder and current patients of a paediatric neurologist or child and adolescent psychiatrist working at CHW were recruited for this treatment study. Ten participants (nine male and one female) completed the program, as two dropped out.

After implementing this modified sensory-motor treatment approach with children with tic disorders, the pilot study findings showed a significant reduction in tic frequency and intensity (158). All ten study participants presented with reduced tic severity following the modified Alert Program® strategies, with the total mean YGTSS score improving from 46.5 to 17.7 post-therapy. Following the intervention, five study participants (study participants 1,2,5,6 and 9) were free of tics. As tics wax and wane, the reason for these participants being tic-free may also be explained by the tics spontaneously waning rather than as a result of therapy (1, 4, 13, 60). The post-test scores from the PTQ reported similar findings in tic reduction, although the

overall change was not significant. The mean pre-post tic severity scores reduced from 49.9 to 31.4. Nine of the 10 study participants showed a reduction in tic severity post-intervention. Study Participant 1 showed an increase in tic scores on the PTQ, which was understood to be a result of different parents completing the measures pre and post-intervention. In contrast, all the other study participants had the PTQ completed by the same parent/carer.

The mechanism by which the Alert Program® is effective, as with other behavioural therapies (57), is unclear. The Alert Program® consists of lessons and activities incorporating both sensory-motor strategies and cognitive approaches. Therefore, this treatment approach is not a 'pure' sensory approach but rather a multi-modal intervention. Other cognitive approaches, such as CBiT, have been effective for children with tic disorders (27, 57, 159, 160). Therefore, it may be the cognitive aspects of the Alert Program® that are more effective than the sensory-motor strategies that are of benefit.

The results from the pilot study showed a significant reduction in tic intensity and frequency, and supported the need for an RCT. However, feedback and insights gained from study participants engaged in our first two studies (Study 1, Chapter 3 and Study 2, Chapter 4) highlighted limitations to current proxy-reported sensory-based measures.

There were functional impairments not captured on the sensory-based measures related to extreme sensitivities to sounds, for example, families not eating meals together due to chewing noises not tolerated by the child, and the sound made from certain fabrics rubbing together, resulting in children experiencing sleep disturbances. Due to these extreme sensitivities to sounds from the study participants' reports, children with tic disorders may experience increased rates of misophonia. This observation was supported by a recent research study

indicating that misophonia could be an underestimated phenomenon for abrupt emotional dysregulation in children with tic disorders (133). This study, published after our clinical observations were made, shows sensory dysregulation experiences in children with tic disorders, particularly sensitivities to sounds, is of global interest and observed not only in our patients.

Additionally, this study recommended that as part of a comprehensive clinical assessment of children with tic disorders, misophonia needs to be assessed (133). Therefore, as these sensory-based measures do not comprehensively capture the extent of the sensory dysregulation problems experienced by the child with tic disorders, particularly relating to misophonia, the impairment cannot be adequately assessed or treated, nor can improvement be appropriately understood.

Another limitation was that these commonly used sensory-based measures capture broader symptoms that overlap with other brain disorders. The behaviours being assessed by the proxy-report sensory-based measures do not relate purely to sensory dysregulation and can be attributed to other neurodevelopmental disorders. For example, item 27 on the CSP2 states: ‘‘my child pursues movement to the point it interferes with daily routine (for example, can’t sit still, fidgets).’’ By scoring this item as frequently occurring, the child may be described as ‘’movement seeking,’’ explained by sensory dysregulation experiences. This same behaviour may be explained through a diagnosis of ADHD (2). This is just one example, but many of the behaviours being rated on this proxy-report sensory-based measure could be explained through a diagnosis of OCD, ASD, ADHD, anxiety, foetal alcohol syndrome, or even childhood trauma (2). The same limitation and examples can also be identified in the SPM assessment measures. Therefore, this brings into question the structural validity of these measures and whether the

underlying factors being assessed by the measures relate purely to sensory dysregulation. Each one of these conditions could possibly require a different treatment response and even different health professionals working with the child and family. It further needs to be acknowledged that ascertaining an appropriate treatment response is problematic without a sensitive, comprehensive, reliable and valid assessment measurement tool.

Parents and children from our first two studies reported sensory dysregulation experiences outside of the items being measured and scored on the current sensory-based measures and did not reflect the impact on daily life. Consequently, we sought to ascertain what properties were being measured using current sensory-based measures to understand why there was a lack of sensitivity in our results. These limitations questioned the measurement design of these sensory measures, the construct they were measuring, their fitness for purpose, and the validity and reliability of these measures. Evidently, there was a need to evaluate the quality of the psychometric properties of current sensory measures, and investigate the possibility of alternative proxy-reported sensory-based measures other than the commonly used measures (Sensory Profile 2 and the Sensory Processing Measure) for use with children with tic disorders.

79.2.3 A Need for a Review of the Current Proxy-Report Sensory-Based Measures used with Paediatric Tic Disorders

A systematic review was undertaken, which aimed to identify all current proxy-reported measures relating to sensory dysregulation in children and adolescents with a neurodevelopmental disorder. Further, this review aimed to comprehensively evaluate the development and psychometric properties of these measures (Study 3, Chapter 5). The researchers had to broaden the scope of the search to include neurodevelopmental disorders

and not only tic disorders in the study population, due to no assessment measures being explicitly identified for children with tic disorders. The Consensus-based Standards for the selection of health Measurement Instruments (COSMIN), a multi-step process, was utilised as the appraisal tool to evaluate the quality of studies, and the quality of psychometric measurement properties of sensory-based measures meeting inclusion criteria for this systematic review (157, 161-163).

Following a systematic search of eleven databases, 6748 articles were screened, 91 full-length articles were reviewed after removing excluded studies, and manual searches were conducted by two reviewers. Twelve proxy-report sensory-based measures were identified from 20 articles. Of the 12 measures, only three provided sufficient data to evaluate content validity, which is the first step of the COSMIN process, and an imperative step to ensure the assessment measure is relevant, comprehensive and comprehensible to the audience completing the measure (164). Thereafter the psychometric properties of measures for internal structure (including structural validity, internal consistency, and cross-cultural validity) (step 2) and the properties of measures for other measurement properties (including reliability, measurement error, criterion validity and hypothesis testing for construct validity including convergent and discriminant validity) (step 3) were evaluated for all twelve measures.

This systematic review identified major issues with the current proxy-report sensory-based measures. None of the twelve measures identified met the criteria for good measurement properties across all ten psychometric properties. Additionally, only three measures (Assessment of Sensory Processing and Executive Function in Childhood (EPYFEI) (165), Child Sensory Profile 2 (CSP2) and the Participation and Sensory Environment Questionnaire-Home (PSEQ-H) (166-168)) published reports on measure development and content validity.

Without evidence that an assessment measure has good content validity, it is impossible to know if it is comprehensive, relevant and comprehendible to the patients completing the questionnaire (156, 157, 162, 163, 169). Therefore according to COSMIN guidelines, assessment measures that do not pass content validity should not be further evaluated in terms of their psychometric properties, as it is not recommended that a measure without good content validity be used in clinical practice (156, 157, 162, 163, 169).

Of the three measures that provided information on the content validity of the tools, only one measure, the Participation and Sensory Environment Questionnaire-Home (PSEQ-H) (166-168), satisfied moderate content validity and moderate-to-high quality for eight of the ten psychometric measurement properties. This measure, although scoring better than the commonly used sensory measures, the Child Sensory Profile 2 and the Sensory Processing Measure, still had limitations for use with children with tic disorders. The items on the PSEQ-H did not reflect all the sensory dysregulation experiences that children with tic disorders and their parents reported through their participation in our first two studies (Study 1, Chapter 3 and Study 3, Chapter 4), particularly concerning misophonia (as discussed in the first finding above). Hence, the PSEQ-H was not comprehensive and relevant enough as a complete proxy-report sensory-based measure for use with children and adolescents with tic disorders.

In summary, the results supported that a proxy-reported sensory-based measure co-designed with young people with tic disorders and their parents was warranted. To address the measurement gaps of sensory dysregulation experiences not currently rated on the existing measures, the new measure needs to have good content validity and psychometric properties across all ten measurement properties. This new proxy-report sensory-based measure needs to be comprehensive enough to assess the broad sensory dysregulation experiences experienced

by all children and young people with tic disorders. The first step of designing a new proxy-report measure according to COSMIN standards is to design the questionnaire with consumers to ensure the questionnaire is comprehensive, comprehensible and relevant to the end user (156, 157). We completed this first of designing a new sensory-based measure through qualitative interviews with young people with tic disorders and their parents to help gain an understanding of their experience of sensory dysregulation and the associated functional impairments (Study 4, Chapter 6). From here, future research will need to address the content validity of the new measure through consultation with consumers and validation testing.

79.2.4 A Broader Scope for the Development of a New Proxy-Report Sensory-Based Measure for Children and Adolescents with Tic Disorders

Subsequently, we sought patient experiences through a qualitative study (Study 4, Chapter 6) completed with sixteen families, young people with tics disorders and their parents to explore their lived experience of sensory dysregulation. This fourth and final study in the thesis is the first step in co-designing a comprehensive, comprehendible, and relevant proxy-reported sensory measure with patients.

An overachieving theme, ‘‘sensory, emotions and tics, it’s a ticking time bomb,’’ emerged from semi-structured interviews with young people with tics and their parents as the higher-order theme (Study 4, Chapter 6). The knowledge gained from these qualitative interviews reiterates the findings from our prevalence study (Study 1, Chapter 3) that children with tic disorders experience difficulties with sensory and emotional dysregulation. These interviews provide greater insight beyond the findings from our prevalence study, in that study participants explained an interplay between sensory and emotional dysregulation that exacerbated tic

expression. Three sub-themes were also identified: 1) we sacrifice and adapt to get daily activities done in the home, 2) my child's experience of the community environment hinder participation, and 3) these sensory preference impact our entire family. Sensory dysregulation experiences specific to tic disorders were identified, such as the impact of excessive temperature, the inability to tolerate the sound of other people chewing, swallowing or scraping of cutlery on crockery, and food intolerances due to seeds in food products.

These tic-specific sensory dysregulation experiences herald the need for a proxy-reported sensory-based measure of sensory dysregulation experiences not assessed in other measures to ensure the sensitivity and specificity required for treatment planning and measuring treatment response. A novel finding from our qualitative study (Study 4 and Chapter 6) highlighted that sensory dysregulation experiences impact the entire family's quality of life and ability to engage in activities, not only the child experiencing the sensory difficulties. Consequently, contrary to current proxy-report sensory-based measures to measure the impact of functional impairment, measurement of the functional deficits or implications on the entire family unit, and not just the child, is needed.

79.3 Implications of the Research Findings

Findings from this research significantly contribute to a comprehensive multidisciplinary assessment and treatment approach for children and adolescents with tic disorders. Children with tic disorders in the presence of comorbidities experience elevated sensory dysregulation and executive dysregulation (Study 1, Chapter 3 and Study 4, Chapter 6). In the interest of a comprehensive assessment of children with tic disorders, is it necessary to evaluate tic severity and premonitory urge, in addition to comorbidities (55), including sensory dysregulation and executive dysfunction.

It is not current practice to assess sensory dysregulation in children with tic disorders and comorbidities, yet 86% of study participants experienced sensory dysregulation experiences (Study 1, Chapter 3). This thesis provides clear evidence that children with tic disorders and comorbidities experience sensory dysregulation. Through interviews with young people with tics and their parents, it is apparent that these sensory problems can significantly affect daily function, participation and quality of life for the child and the entire family unit (Study 4, Chapter 6). These sensory dysregulation experiences result in severe functional impairment, such as being unable to attend school, sports activities or social and community events, travel in a vehicle and attend medical and therapy appointments. Hence, it should be considered that children with tic disorders be assessed and treated by a multidisciplinary team and a multi-modal approach be implemented, which considers sensory dysregulation as part of the assessment and treatment approach.

Regarding the pilot study (Study 2, Chapter 4), this is the first research study to trial the effectiveness of a sensory-based treatment approach with children with tic disorders to reduce tics. This treatment approach is cost-effective, time-effective, child centred and appears feasible. Thus the research studies making up this thesis provide new knowledge into the assessment and treatment of children and adolescents with tic disorders.

79.4 Limitations of the thesis

A limitation of this research was that the recruited participants were a referral cohort from a tertiary paediatric hospital service rather than a population primary care-based. Therefore, there was the potential for severity bias in studies one, two and four. It is, therefore, plausible that a broader study of children with tic disorders may show different rates of sensory dysregulation and executive function difficulties compared with the findings from the prevalence study in

this thesis (Study 1, Chapter 3). However, we are most interested in applying our findings to improve assessment and treatment paradigms, so a treatment-seeking cohort with more severe needs referred to a tertiary children's hospital was deemed suitable.

A further limitation was that a cross-sectional study design was implemented for the prevalence study (Study 1, Chapter 3), which would not capture the variability of tic severity in patients, given that tics wax and wane. A longitudinal study would have assisted in determining dynamic trends in terms of changing sensory dysregulation with the course of the tic pattern and other symptoms. Due to the limitations placed on this research study due to the time frame of the thesis, such a study was not possible, but the finding from our cross-sectional study suggest that a future longitudinal study is warranted.

Furthermore, in relation to Study 2 (Chapter 4), when pilot testing the modified version of the Alert Program® with children with tics, a one-month follow-up period is too short a time frame to confirm whether the benefit identified by the treatment approach is sustained when considering the waxing and waning nature of tics. This is a limitation of the pilot study (Study 2, Chapter 4) making up this thesis. Additionally, as an open-label study design that is open to bias was implemented, future studies must include a control arm. Further research into the long-term effects of this intervention is needed.

In relation to the systematic review, although the search strategy included search terms for all ten psychometric properties being evaluated according to COSMIN, no studies were identified for any of the twelve measures identified on responsiveness. Whilst an additional search strategy specifically focused on responsiveness was not conducted, it is unlikely that the findings from such a study would influence the salient outcomes for this cohort. An additional

systematic review to specifically identify all studies that have used any of the twelve identified sensory-based measures in intervention studies to evaluate the responsiveness of these sensory-based measures may be relevant for other neurodevelopmental conditions. This would require a meta-analysis of the pre-/post-test data to determine these sensory-base measures' responsiveness. This additional systematic review was outside the scope of this thesis and is therefore recommended for future research.

Through the knowledge gained from the systematic review (Study 3, Chapter 5) relating to the psychometric properties of the proxy-reported sensory-based measures, it was identified that the PSEQ-H was the only proxy-reported sensory-based measure with adequate content validity. As the proxy-report sensory-based measures used in Study 1 (Chapter 3) were the Child Sensory Profile 2 and the Sensory Processing Measure, which were shown to have inadequate or no content validity, respectively, it would be recommended that this prevalence study be repeated using a sensory-based measure with good psychometric properties, such as the PSEQ-H. In addition, it would be recommended that once a valid and reliable sensory-based measure is designed to evaluate sensory dysregulation in children with tic disorders, the prevalence of sensory dysregulation symptoms be re-assessed.

Finally, although through these research studies, we have identified that sensory dysregulation occurs in children with tic disorders, the research further identified that proxy-report sensory-based measures that clinicians and researchers use to evaluate sensory dysregulation are imperfect. These findings together create a 'discordance' and 'dichotomy' that needs to be corrected. Clinicians and researchers working in the field of paediatric tic disorders may be better supported through the development of a new proxy-report sensory-based measure to allow for accurate and comprehensive assessment and monitoring of change in patients.

Additionally, a child-centred, cost-effective, efficient intervention that completes existing practice is needed to support children with tic disorders and comorbidities where current therapy is ineffective or insufficient.

79.5 Recommendations for Future Research and Practice

Eighty-one percent of the study participants in the prevalence study had a diagnosis of TS rather than tic disorder. Future studies should compare the different tic diagnoses (i.e., chronic vocal tic, chronic motor tic, TS etc.). There is also the possibility that sensory dysregulation, premonitory urge, and tic severity are linked in terms of the sensory nature of these experiences. Therefore, future studies should address whether the premonitory urge (using the Premonitory Urge for Tics Scale) and sensory dysregulation are associated.

These research findings support future research focusing on co-designing a relevant, comprehensive and comprehendible proxy-report sensory-based measure specific enough to pick up the change in sensory dysregulation in children and young people with tic disorders. This assessment measurement also needs to evaluate the impact of sensory dysfunction and impairment on the entire family unit. The qualitative study (Study 4, Chapter 6) was phase 1 of developing a new sensory-based assessment measure. The qualitative interviews allowed for the generation of items for the new measure. Future research needs to focus on Phase 2 and Phase 3 of the development of this assessment measure (Figure 7.2). Phase 2 requires consultation with consumers to ensure content validity requirements of the proposed sensory-based assessment measure are met, according to COSMIN guidelines (156, 157, 164, 169). Thereafter, Phase 3 will be implemented to ensure validation testing of the psychometric properties of the new assessment measure before it can be used in clinical and research practices.

In terms of the assessment of sensory-based measures, only proxy-report measures were focused on in this thesis. The complexity and volume of measures precluded appraisal of clinician-and teacher-rated sensory-based measures for children and adolescents, and were outside the scope of the systematic review. As validated, sensitive, reliable, and responsive clinician-, teacher-, patient-, and proxy-reported outcome measures to assess treatment efficiency are necessary for clinical use in sensory dysregulation, future research needs to focus on a systematic review of clinician- and teacher-reported sensory based measures using COSMIN as the appraisal tool.

Due to the significant improvement in tic reduction in children with tic disorders after the implementation of a modified version of the Alert Program®, there is a need for further research into the effectiveness of this treatment approach in the form of a randomised controlled trial. With the development of a valid, reliable, fit-for-purpose proxy-report sensory-based assessment measure, this tool can be utilised in this RCT.

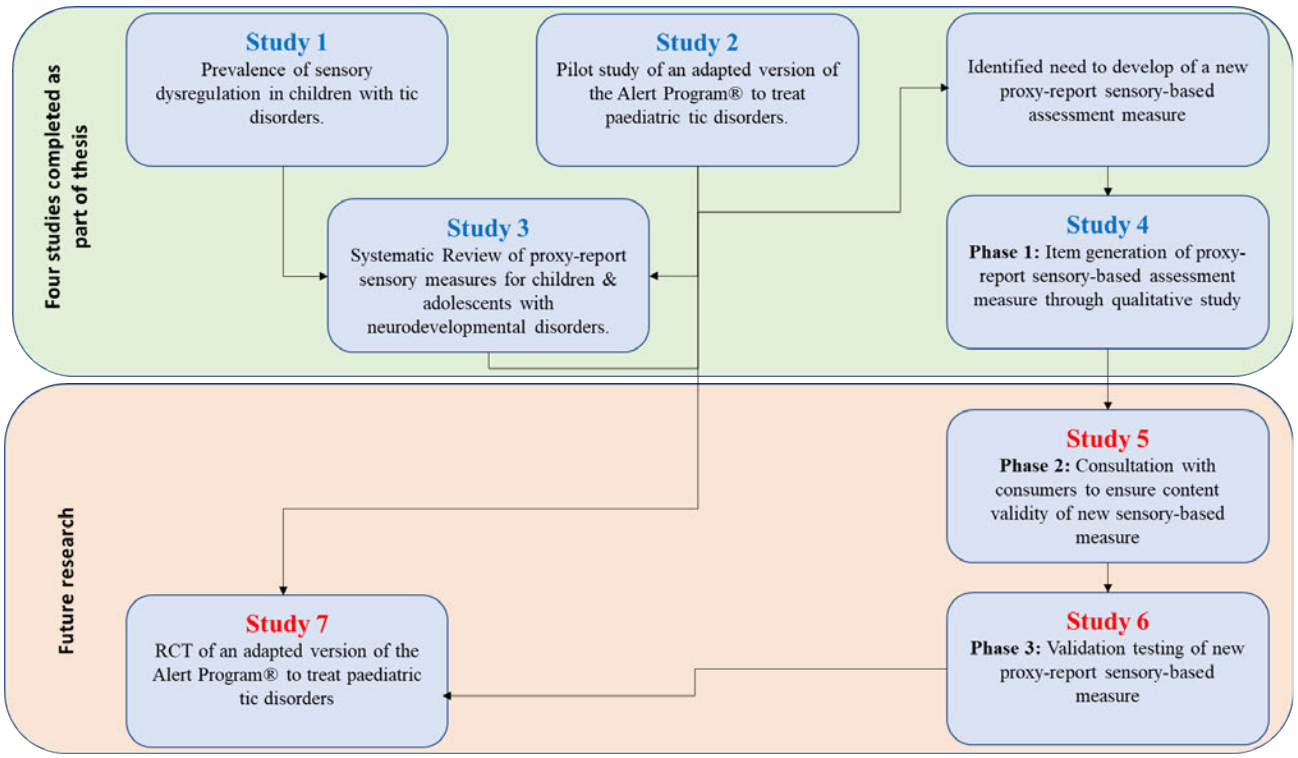

Figure 7.2 Flowchart of research studies conducted as part of this thesis and recommended future research as a result of the findings from the completed studies. Validation testing of the new proxy-report sensory-based measure (Study 6) would include structural validity, internal consistency, reliability, measurement error, criterion validity, and hypothesis testing for construct validity and responsiveness(169). Responsiveness of the measure would need to be established prior to the measure being able to be used in Study 7 as an outcome measure. As the newly developed sensory-based measure would be implemented with the population it was designed for, cross-cultural validity of the measure at this stage would not be required but would be a future consideration should there be a need to use this measure with a different population group.

79.6 Concluding Statement

From the four research studies conducted as part of this thesis, our research offers an understanding of the prevalence and impact of sensory dysregulation experiences in children with tic disorders. With sensory dysregulation being significantly prevalent in children with tic disorders and comorbidities, and these sensory dysregulation experiences' impact on the entire family unit, there is a need for comprehensive assessed and treated which considers sensory dysregulation. Furthermore, it is evident that there is a need for new sensory assessment measures that we are co-designing with patients with tics and their families. Assessment of children with tic disorders needs to be comprehensive and focus not only on tic severity, the premonitory urge and comorbidities but also include an understanding of sensory dysregulation and executive function. The significant improvements identified in tic severity following the pilot study of a sensory-based treatment show that sensory-motor intervention may bridge the treatment gap for young children with tics.

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APPENDICES

Appendix I: Ethics Approvals

The Sydney children's Hospitals Network
care, advocacy, research, education

Contact for this correspondence:
Research Ethics Office
Research Ethics Administration Assistant
Phone: (02) 9845 1253
Facsimile: (02) 9845 1317
Email: SCHN-ethics@health.nsw.gov.au

Corner Hawkesbury Road
and Hainsworth Street
Locked Bag 4001
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DX 8213 Parramatta
Tel +61 2 9845 0000
Fax +61 2 9845 3489
http://www.schn.health.nsw.gov.au/
ABN 53 188 579 090

22/02/2017

Miss Nicolette de Kock
Senior Occupational Therapist
Psychological Medicine & Occupational Therapy Department
The Children's Hospital at Westmead

Dear Ms de Kock,

HREC reference number:	LNR/17/SCHN/8
Project title:	Prevalence of sensory symptoms in children with tic disorders
Site/s	One – The Children's Hospital at Westmead

Thank you for submitting the above project for single ethical and scientific review. This project was considered by the Sydney Children's Hospitals Network Human Research Ethics Committee's Executive Committee ("the Committee") at its meeting on 19th January 2017, and twice thereafter by the Executive Officer of the SCHN HREC at their meetings of 13 February 2017 and 22 February 2017.

This HREC has been accredited by the NSW Department of Health as a lead HREC under the model for single ethical and scientific review, and by the National Health and Medical Research Council as a certified committee in the review of multi-centre clinical research projects.

This HREC is constituted and operates in accordance with the National Health and Medical Research Council's National Statement on Ethical Conduct in Human Research and CPMP/ICH Note for Guidance on Good Clinical Practice.

I am pleased to advise that the Committee has granted ethical approval of this research project. Your approval is valid for **three (3) years**, effective the date of this letter.

This application has been assessed in accordance with, and meets the requirements of the National Statement on Ethical Conduct in Human Research (2007).

The documents approved and/or noted by the SCHN Human Research Ethics Committee are;

Title	Version	Date of Document	Date of submission
LNR Application – AU/6/E21B215	-	09/01/2017	
Clinician Questionnaire 2016	v1		13/02/2017
Invitation Letter to Potential Participants	v2	13/02/2017	
Information Sheet for Parents	v4	15/02/2017	
Staff Invitation	v2	13/02/2017	
Protocol	v2	February 2017	
PedsQL-Core-UserAgreement	v3		31/01/2017
PedsQL-4 0-Core-PYC_AU4 0_eng-AU 5-7 years	-	28/01/2013	
Nicolette De Kock CV			10/01/2017
Parent Report Measures for Children and Adolescents SDQ(P)04-10	-		10/02/2017
Sensory Processing Measure Autoscore Form	-	2007	10/01/2017
Child Sensory Profile 2 Caregiver Questionnaire 3:0 – 14:11 years -	-	-	10/01/2017
Yale Global Tic Severity Scale		1992	10/01/2017

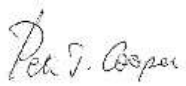
Please note the following conditions of approval:

1. The Coordinating Investigator will immediately report anything which may warrant review of ethical approval of the project in accordance with the SCHN adverse event reporting policy.
2. All proposed changes to the research protocol, including the conduct of the research, changes to site or personnel, or an extension to HREC approval, are to be provided to the HREC or its delegate for review before those changes can take effect.
3. The HREC will be notified, giving reasons, if the project is discontinued at a site before the expected date of completion.
4. The co-ordinating investigator will provide an annual report to the HREC on the anniversary of this approval letter, and a final report on completion of the study.
5. Your approval is valid for three (3) years from the date of the final approval letter. If your project extends beyond that three year period and you are still actively recruiting you will be required to resubmit your application incorporating any amendments within six (6) months of that approval expiry date. If your project is in follow up on, or analysis, please submit an application for amendment to extend the approval period. Ethics approval can be extended for a period of twelve (12) months at a time.
6. In the event of a project not having commenced within 12 months of its approval, the approval will lapse and reapplication to the HREC will be required.

You are reminded that this letter constitutes **ethical approval only**. You must not commence this research project at a site until separate authorisation from the Chief Executive or delegate of that site has been obtained. A copy of this letter must be forwarded to all site investigators for submission to the relevant Research Governance Officer.

The HREC wishes you every success in your research.

Yours faithfully



Dr Peter Cooper
Chair, Sydney Children's Hospitals Network Human Research Ethics Committee

The Sydney children's Hospitals Network

care, advocacy, research, education

Contact for this correspondence:
Research and Development
Name: Geraldine Bicol
Phone: (02) 9845 3011
Facsimile: (02) 9845 1317
Email: Geraldine.Bicol@health.nsw.gov.au

4 May 2017

Nicolette de Kock
Senior Occupational Therapist
Department of Psychological Medicine and Occupational Therapy
The Children's Hospital at Westmead

Site Authorisation Letter

Dear Nicolette,

HREC reference number: LNR/17/SCHN/8

SSA reference number: LNRSSA/17/SCHN/15

Project title: **Prevalence of Sensory Symptoms in Children with Tic Disorders**

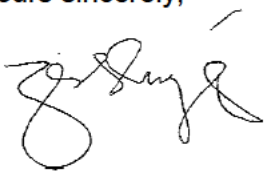
Site: **The Children's Hospital at Westmead**

Thank you for submitting an application for authorisation of this project. I am pleased to inform you that authorisation has been granted for this study to take place at the above site.

The following conditions apply to this research project. These are additional to those conditions imposed by the Human Research Ethics Committee that granted ethical approval:

1. Please advise us of the date when the project starts at this site.
2. Proposed amendments to the research protocol or conduct of the research which may affect the ethical acceptability of the project, and which are submitted to the lead HREC for review, are copied to the research governance officer.
3. Proposed amendments to the research protocol or conduct of the research which may affect the ongoing site acceptability of the project are to be submitted to the research governance officer.

Yours sincerely,



Geraldine Bicol
Research Governance Officer

Locked Bag 4001
Westmead NSW 2145
Sydney Australia

Tel 61 2 9845 0000
Fax 61 2 9845 3489
www.schn.health.nsw.gov.au

ABN 53 188 579 090

DX 8213 Parramatta

Section 1.02 Identification of the Questionnaire

Title of the Questionnaire:	PedsQL™ (Pediatric Quality of Life Inventory™)
Author:	James W. Varni
Owner:	James W. Varni
Copyright notice	Copyright © 1998 JW Varni, Ph.D. All rights reserved.
References:	See Appendix 2

Article 2. RIGHTS TO USE

Section 2.01 Context of the Use of the Questionnaire

The User undertakes to only use the Questionnaire in the context of the Study as defined hereafter.
[Tick the box and complete the corresponding fields]

☐ *individual clinical practice (please go directly to section 2.02)*

Planned term of use: .....

Number of patients expected: .....

☒ *clinical project or study*

Title:	Prevalence of Sensory Processing Differences in children with tic disorders in an Australian Population .....
Study/protocol reference:	.....
Disease or condition:	Tic Disorders is the focus of the study but cohort groups will include: children with ASD and Epilepsy .....
Type of research:	☐ clinical trial : ☐Phase II / ☐Phase III ☐ epidemiologic/observational ☒ other: ...Parent completed surveys and questionnaires .....
Questionnaire used as primary end point:	☐ yes ☒ no
Number of patients expected:	280.....
Number of submissions to the Questionnaire for each patient:	1 per patient.....
Term of clinical follow-up for each patient:	None required, only data collected on initial symptoms .....
Planned term for project:	start (month/year): March 2017..... end (month/year): Dec 2017 .....
Mode of Administration:	☒ paper ☐ electronic
If electronic administration, please indicate mode of data collection:	☐ Hand held device – specify device: ..... ☐ Interactive Voice Response (IVR) – specify: ..... ☐ Web - specify website: ..... ☐ Digital Pen - specify device: ..... ☐ Tablet - specify device: ..... ☐ other - specify: .....

Article 5. **TERRITORIES AND LANGUAGES**

MRT transfers the Limited Rights to use the Questionnaire on the following territories and in the languages indicated in the table below:

Language:	*For use in the following country*	*Language:*	*For use in the following country*	*Language:*	*For use in the following country*
English	*Australia*				
-	-				

Article 6. **PRICE AND PAYMENT TERMS**

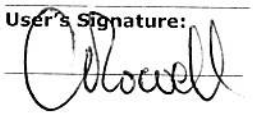
The User undertakes in relation to MRT to pay the price owed in return for the availability of the Questionnaire, according to the prices set out in Appendix 3, depending on the languages requested and the costs of using the Questionnaire, in accordance with the terms and conditions described in section 6.02 of the General Terms included in Appendix 1.

Agreed and acknowledged by:

User's Name:

Prof. Chris Cowell

User's Signature:



User's Title:

Director of Research

Date:

1/3/2017

The Sydney children's Hospitals Network
care, advocacy, research, education

Contact for this correspondence:
Research Ethics Office
Research Ethics Administration Assistant
Phone: (02) 9845 1253
Facsimile: (02) 9845 1317
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ABN 53 188 579 090

24 April 2017

Miss Nicolette de Kock
Psychological Medicine, Neurology and Occupational Therapy
The Children's Hospital at Westmead

Dear Miss de Kock,

HREC Reference:	**LNR/17/SCHN/8**
Project title:	**Prevalence of sensory symptoms in children with tic disorders**
Site/s:	**The Children's Hospital at Westmead**

I acknowledge receipt of your project amendment submitted 07 March 2017, requesting approval for:

1. The Behaviour Rating Inventory of Executive Function (BRIEF) Parent Form Questionnaire be included into the research study and be an addition to the questionnaire completed by the participants in the study.

2. The Parent Forms of the BRIEF each contain 86 items that measure different aspects of executive function. Eight clinical scales (Inhibit, Shift, Emotional Control, Initiate, Working Memory, Plan/Organize, Organization of Materials, Monitor) and two validity scales (Inconsistency and Negativity) give the clinician a well-rounded picture of the behaviour of the child being rated. The additional questionnaire is expected to add 10 to 15 minutes additional time to score.

3. Add questionnaire PedsQL (8-12 years) as it was omitted in original application

The amendment/s was reviewed at the meeting of the Executive Committee of the Sydney Children's Hospitals Network Human Research Ethics Committee (SCHN HREC) at its meeting held on 15 March 2017 and subsequently out of session by the Executive of the SCHN HREC on 24 April 2017.

I am pleased to advise that, following receipt of the further information required on 18 April 2017, the documents reviewed and approved were:

Documents Reviewed	*Version*	*Date*
Amendment Form	-	27 February 2017
Email amendment request to review extra document	-	09 March 2017
BRIEF Parent form	-	Rec'd 07 March 2017
Invitation Letter to Potential Participants	V3	27 February 2017
PedsQL Parent Report for Children (ages 8-12)	V4.0	rec'd 07 March 2017
Staff Invitation	V3	27 February 2017
Prevalence cross sectional study of SPD in children with tic disorders	V3	7 March 2017
Email response to request for further information	-	18 April 2017
Parent Information Sheet	V6	18 April 2017

This lead HREC is constituted and operates in accordance with the National Health and Medical Research Council's *National Statement on Ethical Conduct in Research Involving Humans* and the *CPMP/ICH Note for Guidance on Good Clinical Practice*.

This letter constitutes ethics amendment approval ONLY. A copy of this letter must be forwarded to the Research Governance Officer at each site for governance approval.

This application has been assessed in accordance with, and meets the requirements of the National Statement on Ethical Conduct in Human Research (2007).

Should you require any further information, please do not hesitate to contact the Research Ethics Office at SCHN-ethics@health.nsw.gov.au or on (02) 9845 1253.

Yours sincerely,

[REDACTION]

Associate Professor Sarah Garnett
Chair, Sydney Children's Hospitals Network Human Research Ethics Committee
Sydney Children's Hospitals Network Human Research Ethics Committee

PARENT INFORMATION SHEET

Prevalence of sensory symptoms in children with tic disorders.

Thank you for taking the time to read this Information Sheet. We would like you to consider participating in a research study being undertaken by the Neurology and Psychological Medicine Departments at The Children's Hospital at Westmead in conjunction with The University of Sydney.

This study is about comparing children's responses to sensory stimulation from their everyday environment. We are comparing the responses of two different groups of children. The study involves parents answering questions and there is no child involvement in this study at all.

These groups are:

1. Parents whose children have a diagnosis of tic disorders,
2. A control group. The control group is made up of parents who are staff members of the Children's Hospital at Westmead and have children with no known or suspected neurodevelopmental, psychiatric or mental health diagnosis.

The aim of the study is to understand if children with tic disorders have increased sensory symptoms or sensitivities. Understanding the presentation of sensory symptoms children with tic disorders may have compared with other groups of children, will allow us to improve our assessment process and our treatment for children with tic disorders. The study also allows us to assess the effectiveness of current sensory screener questionnaires that are used commonly.

Please read this Information Sheet carefully. You can ask us questions about anything at any time.

The following pages tell you about the project. It explains to you clearly and openly all the steps and procedures of the project. The information is to help you to decide whether or not you would like to take part in the research.

Participation in this research project is voluntary. If you don't want to take part, you don't have to. You can withdraw from the project at any time without explanation. Withdrawal from the study at any time during the research project will not in any way affect your own, or your child's future health care or services received from The Children's Hospital at Westmead.

This study only requires parent participation through answering questionnaires and by participating you are not actively involving your child in any the study.

Investigator:

Ms Nicolette de Kock, Occupational Therapist and PhD student, Psychological Medicine at The Children's Hospital at Westmead and The University of Sydney. ph.(02) 9845 2005 email nicolette.dekock@health.nsw.gov.au.

Supervisors:
Professor Russell Dale, Department of Neurology at The Children's Hospital at Westmead
Dr Paula Bray Occupational Therapy Department at The Children's Hospital at Westmead

What is the study about?
Children with tic disorders can often have sensory symptoms before the tics occur or many have increased sensory sensitive to sensory experiences. This study aims to find out how common it is for children with tic disorders to experience sensory symptoms compared with other groups of children. Research in this area to date has only looked at using a short questionnaire with children with Tourette's in Canada in 2012. A study has not been carried out with an Australian population of children with tics nor has a more comprehensive study been done.

The information obtained from this study will help us improve our assessment of children with tic disorders. The outcomes of this research will also be used as part of a PhD (Doctor of Philosophy) degree thesis of the Principal Investigator.

What will this study do?
The study allows us to ensure that the most accurate screening questionnaire is used to assess the sensory sensitivities in children with tic disorders as part of an improved and holist assessment approach. This in turn should also assist in aiding our treatment approach and understanding and so improving our quality of care.

We also hope that the information gained from this study will assist with children with tic disorders to be referred to occupational therapists as part of the treating team for a comprehensive and multi-disciplinary team approach to the child's care should this be required, as this is not currently common practice.

What will the study involve?
If you agree to participate, you will be asked to complete 5 questionnaires that ask questions about your child's:

- sensory preferences,
- physical, emotional, social, and school functioning in the past one month
- behavioural responses.

We anticipate that the questionnaires together will take about a total of 40 to 45 minutes to complete. The aim would be that the questionnaires be completed in the waiting room before or after attending your child's pre-planned medical appointment.

The questionnaires which you are being asked to complete are the:

1. A Sensory Processing Questionnaire
2. A Sensory Profile 2 Questionnaire
3. The Peds Quality of Life™
4. Strengths and Difficulties Questionnaire (SDQ)
5. Behaviour Rating Inventory of Executive Function (BRIEF) Parent Form

Once completed, we request that you place the questionnaires in the envelope provided and return it sealed to the Principal Investigator who would be present in your waiting area. Should the questionnaire not be completed in the duration that the participant is at the hospital, a prepaid envelope will be included to allow for the completed questionnaires to be posted back to the Principal Investigator.

For the Principal Investigator to understand your child's diagnosis we seek your permission to access your child's medical records. This will only be for the purpose of understanding your child's diagnosis and the medication and treatment they are receiving. All information will be classified and any data published on the overall participant's scores will be non-identifiable. Participant's data will be re-identifiable as a master sheet with identifying data codes will be kept by the Principal Investigator which links the non-identifiable data to the client. Only the researchers in this study will have access to the master sheet with the identifying code. Therefore it would only be the researchers who could re-identify participant's data in the study.

If you have any questions that you would like answered before you complete the questionnaire, you can approach the Principal Investigator in person as they will be on hand in the waiting room to assist. Should you have any questions following the completion of the questionnaires, please contact the Principal Investigator Nicolette de Kock on (02) 9845 2005.

No further commitments are required once the questionnaires are completed. The Principal Investigator will inform your medical specialist of the results of the questionnaires and they will make any necessary recommendations as required.

Who can participate in the study?

You are being asked to participate in this study because you have a child aged between 5 and 12 years who attends appointments through either the Department of Neurology or Psychological Medicine at The Children's Hospital at Westmead. Parents recruited in the study also have a child with a confirmed diagnosis of one of the following diagnosis: tic disorders, epilepsy, or autism spectrum disorder.

Staff members of The Children's Hospital at Westmead will be invited to participate if they have children between the ages of 5 and 12 years old with no known diagnosis to make the control group.

Are there any benefits for my child participating in the study?

We do not anticipate there to be any direct benefits to you or your child as a result of participating in this study. We do hope that the results will help us to understand more about the sensory experiences of children with tic disorders, which may then help us to improve our clinical practices.

Are there any risks associated with this study?
We do not anticipate there to be any risks associated with participating in this study. The questions asked however may cause you to think about issues that might upset you. If you do feel uncomfortable answering some questions or distressed, please contact the Principal Investigator so that we can discuss the situation, and where necessary, refer you to an appropriate support service. If the Principal Investigator is unavailable and you would like to speak to someone please contact Lifeline on 131114.

What are my alternatives to taking part in this project?
You do not have to take part if you do not want to. If you decide to take part and later change your mind, you are free to withdraw from the project at any stage. Doing so will not affect your relationship with, or the care your child receives at The Children's Hospital at Westmead. You also do not need to provide a reason for your withdrawal from the study.

What will be done to make sure my child's information is confidential?
Confidentiality and anonymity will be preserved in this study. All information collected as part of this study will remain confidential and will only be used for the purposes of this research project, unless as required by law. Any documents that contain identifiable information will be kept securely in a locked filing cabinet. On completion of the questionnaires you will be assigned a study identification number that will be used when transferring information into the database. Paper records will be kept securely for 5 years before being destroyed.

It is intended that the outcomes of this research will also be presented in academic journals and at conferences. At no time during those presentations will you or your child's details be identifiable. Information will only be reported as group data.

Other information

If you have any questions about this study, please do not hesitate to discuss them with the Principal Investigator, Nicolette de Kock ph. 9845 2005 email nicolette.dekock@health.nsw.gov.au.

This project has been approved by the Sydney Children's Hospitals Network Human Research Ethics Committee. If you have any concerns or complaints about the conduct of this study, please do not hesitate to contact the Executive Officer of the Ethics Committee (02 9845 3066) or via email SCHN-ethics@health.nsw.gov.au and quote approval number LNR/17/SCHN/8

This Information Sheet is for you to keep.

Consent Form – Parent / Guardian

Title: **Prevalence of Sensory Differences in Children with Tic Disorders in an Australian Population**

Short Title: **Sensory Differences in Children with Tic Disorders**

Protocol Number:

Project Sponsor: **The Children's Hospital at Westmead**

Coordinating Principal Investigator: **Nicolette de Kock, 02 9845 3369**

Associate Investigator: **Prof. Russell Dale, The Children's Hospital at Westmead, 02 9845 3404**
Dr. Paula Bray, The Children's Hospital at Westmead, 02 9845 3369
Chris Hardwick, The Children's Hospital at Westmead, 02 9845 2005
Iain Perks, The Children's Hospital at Westmead,

Declaration by Parent / Guardian

I have read the Participant Information Sheet or someone has read it to me in a language that I understand.

I understand the purposes, my required involvement, the procedures and risks of the research described in the project.

I have had an opportunity to ask questions and I am satisfied with the answers I have received.

I consent to engage in the study. I understand that my involvement in the study involves the completion of questionnaires relating to my child.

I give permission for research to review my child's medical records for the purpose of understanding my child's medical diagnosis for the purpose of research. I understand that the information obtained through the questionnaires and medical records will remain confidential and only be used for the purpose of research.

I freely agree to participate in this research project as described and understand that I am free to withdraw at any time during the research project without affecting my own, or my child's future health care.

I understand that I will be given a signed copy of this document to keep.

Name of Child (please print): _____

Signature of Child: _____ Date: _____

Name of Parent / Guardian (please print): _____

Signature of Parent / Guardian: _____ Date: _____

Name of Witness* to Parent / Guardian's Signature (please print): _____

Signature of Witness: _____ Date: _____

* The Witness is not to be the investigator, a member of the study team or their delegate. In the event that an interpreter is used, the interpreter may not act as a witness to the consent process. Witnesses must be over 18 years of age.

11.5.2017

Dear CHW Staff Member

Re: Invitation to Participate in a Research Study (Prevalence of sensory symptoms in children with tic disorders)

Thank you for taking the time to consider your participation in this research study being undertaken by the Neurology and Psychological Medicine Departments at The Children's Hospital at Westmead in conjunction with The University of Sydney. The purpose of the study is to understand if children with tic disorders have increased sensory symptoms or sensitivities. This information will help guide our treatment approaches.

You are being invited to be a part of the control group for this study. The study involves parents answering questions and there is no child involvement in this study at all. You will be asked to complete 5 questionnaires that ask questions about your child's sensory preferences, physical, emotional, social, and school functioning and their behavioural responses. Included with this invitation letter is a Participant Information Sheet which will provide more information about the research and your invited role in the study.

Should you be willing to participate in the study, please contact Nicolette de Kock, so that you can be provided with the questionnaires to complete. It will also be an opportunity to have any questions you may have related to the study answered. Questionnaires can be completed at home and returned via a pre-paid envelope.

There are no implications should you not wish to participate in the study.

If you have any questions about this study, please do not hesitate to discuss them with the Principal Investigator, Nicolette de Kock ph. 9845 2005 email nicolette.dekock@health.nsw.gov.au.

Thank you for your time.

Yours Sincerely

Nicolette de Kock
Senior Occupational therapist
Psychological Medicine and Occupational Therapy Departments
The Children's Hospital at Westmead

The Sydney children's Hospitals Network
care, advocacy, research, education

Contact for this correspondence:
Research Ethics Office
Research Ethics Administration Assistant
Phone: (02) 9845 1253
Facsimile: (02) 9845 1317
Email: SCHN-ethics@health.nsw.gov.au

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ABN 53 188 579 090

25 March 2020

Ms Nicolette Soler
Department of Paediatric Neurology& Psychological Medicine
The Children's Hospital at Westmead

Dear Ms Soler,

HREC Reference: **2020/ETH00132**

Project title: **Development and Trial of a Sensory Assessment Measure for Children & Young People with tic disorders**

Sites: **The Children's Hospital at Westmead**

Thank you for submitting the above project for single ethical and scientific review. This project was first considered by the Sydney Children's Hospitals Network Human Research Ethics Committee ("the Committee") at its meeting **21 February 2020** and subsequently by the **Executive of SCHN HREC** and HREC Delegates on the **23 March 2020**.

The HREC has been accredited by the NSW Department of Health as a lead HREC under the model for single ethical and scientific review, and by the National Health and Medical Research Council as a certified committee in the review of multi-centre clinical research projects.

This HREC is constituted and operates in accordance with the National Health and Medical Research Council's *National Statement on Ethical Conduct in Human Research* and *CPMP/ICH Note for Guidance on Good Clinical Practice.*

I am pleased to advise that the Committee has granted ethical approval of this research project. Your approval is valid for five (5) years, effective the date of this letter.

This application has been assessed in accordance with, and meets the requirements of the National Statement on Ethical Conduct in Human Research (2007).

The documents reviewed and approved by the Committee are:

Document	*Version*	*Date*
Protocol	V2	23 Mar 2020

Document	*Version*	*Date*
REGIS Project Registration	-	Rec'd 29 Jan 2020
Parent Follow up Phone Script	V2	11 Mar 2020
Family invitation letter Phase 1	V2	11 Mar 2020
Parent PIS Phase 1	V2	11 Mar 2020
Young person PIS phase 1	V1	08 Jan 2020
Parent and young person Consent Form- Phase 1	V2	11 Mar 2020
Parent semi-structured interview questions- Phase 1	V2	11 Mar 2020
Young person semi-structured interview questions- Phase 1	V2	11 Mar 2020
Parent semi-structured interview ADL poster prompt Phase 1	V2	11 Mar 2020
Young person semi-structured interview ADL poster prompt Phase	V2	11 Mar 2020
Parent semi-structured interview sensory poster prompt phase 1	V1	08 Jan 2020
Young person semi-structured interview sensory poster prompt phase 1	V1	28 Oct 2019
Demographic survey Phase 1	V1	08 Jan 2020
Psychological distress plan phase 1, 2 & 3	V1	08 Jan 2020
Consumer invitation letter phase 2	V2	11 Mar 2020
Consumer Information Sheet	V2	11 Mar 2020
Consumer phone script phase 2	V2	11 Mar 2020
Consumer advertisement of invitation letter for TSAA	V2	11 Mar 2020
Parent Invitation letter Phase 3	V2	11 Mar 2020
Parent PIS Phase 3	V2	11 Mar 2020
Parent Consent Form Phase 3	V2	11 Mar 2020
Parent Follow up Phone Script Phase 3	V2	11 Mar 2020
Sensory Profile 2 - standardised measure	-	Rec'd 29 Jan 2020
SDQ- English - standardised measure	-	Rec'd 29 Jan 2020

Document	*Version*	*Date*
Brief 2 Parent form - standardised measure	-	Rec'd 29 Jan 2020
Peds QL4-OPC-5-7 yrs - standardised measure	-	Rec'd 29 Jan 2020
Peds QL4-OPC-8-12 yrs - standardised measure	-	Rec'd 29 Jan 2020
Demographic survey phase 3	V1	08 Jan 2020
YGTSS- Yale Global Tic Severity Scale- standardised measure	-	Rec'd 29 Jan 2020
Advertisment of invitation letter for Bandage Bear	V2	11 Mar 2020
SCHN Response to Committee 2020-ETH00132	-	23 Mar 2020
Email regarding updated documents	-	23 Mar 2020
HREA	V2	28 Feb 2020

Please note the following conditions of approval:

1. The Coordinating Investigator will immediately report anything which may warrant review of ethical approval of the project in accordance with the SCHN adverse event reporting policy.

2. All proposed changes to the research protocol, including the conduct of the research, changes to site or personnel, or an extension to HREC approval, are to be provided to the HREC or its delegate for review before those changes can take effect.

3. The HREC will be notified, giving reasons, if the project is discontinued at a site before the expected date of completion.

4. The co-ordinating investigator will provide an annual report to the HREC on the anniversary of this approval letter, and a final report on completion of the study.

5. Your approval is valid for five (5) years from the date of the final approval letter. If your project extends beyond that five year period and you are still actively recruiting you will be required to resubmit your application incorporating any amendments within six (6) months of that approval expiry date. If your project is in follow up on, or analysis, please submit and application for amendment to extend the approval period. Ethics approval can be extended for a period of twelve (12) months at a time.

6. In the event of a project **not having commenced** within 12 months of its approval, the approval will lapse and reapplication to the HREC will be required.

Should you have any queries about the HREC's consideration of your project please contact the Ethics Administration Assistant on (02) 9845 1253.

You are reminded that this letter constitutes ethical approval only. You must not commence this research project at a site until separate authorisation from the Chief Executive or delegate of that site has been obtained. A copy of this letter must be forwarded to all site investigators for submission to the relevant Research Governance Officer.

The SCHN HREC wishes you every success in your research.

Yours faithfully

[REDACTION]

Associate Professor Sarah Garnett
Chair, Sydney Children's Hospitals Network Human Research Ethics Committee
Sydney Children's Hospitals Network Human Research Ethics Committee

***NB: All clinical trials must now be registered on a publicly accessible registry such as the Australian New Zealand Clinical Trials Registry. For further information please go to www.anzctr.org.au*. Please provide this office with a copy of your registration number for our records if you have not already done so.**

Contact for this correspondence:
Name: Amelia Assareh
Phone: (02) 9845 3011
Facsimile: (02) 9845 1317
Governance inbox: SCHN-Governance@health.nsw.gov.au

3 April 2020

Ms Nicolette Soler
Department of Paediatric Neurology& Psychological Medicine
The Children's Hospital at Westmead

Site Authorisation Letter

Dear Ms Soler,

HREC reference number: 2020/ETH00132

SSA reference number: 2020/STE00307

Project title: *Development and Trial of a Sensory Assessment Measure for Children & Young People with tic disorders*

Site/s: **The Children's Hospital at Westmead**

Thank you for submitting an application for authorisation of this project. I am pleased to inform you that authorisation is granted for this study to take place at the above site.

The following conditions apply to this research project. These are additional to those conditions imposed by the Human Research Ethics Committee that granted ethical approval. ***Site authorisation may be withdrawn if these conditions are not met.***

1. Please advise us, via email, the **date when the project starts** at this site.

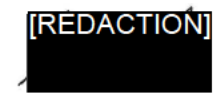
2. Proposed **amendments** to the research protocol or conduct of the research which may affect the **ethical acceptability** of the project, and which is submitted to the lead HREC for review, are submitted to the Research Governance Officer.

3. Proposed **amendments** to the research protocol or conduct of the research which may affect the ongoing **site acceptability** of the project are to be submitted to the research governance officer.

4. A copy of the **annual report** submitted to the lead HREC must be provided to this office **after receipt of HREC acknowledgement.**

All site post-authorisation reports and amendment applications should be sent at first instance to the SCHN-Governance inbox. Please visit our intranet or internet site for more information.

Yours sincerely,



Amelia Assareh (PhD)
Research Governance Officer

From:	no_reply@regis.health.nsw.gov.au
To:	Nicolette Soler (SCHN)
Cc:	SCHN-Ethics
Subject:	2020/ETH00132: Notification of an amendment to a research study - General Amendment (97286) - Approved
Date:	Wednesday, 22 June 2022 10:51:11 AM
Attachments:	097286_Figure sumary of themes and sub themes V1 16.06.2022 - HREC comments.pptx.pptx

Date of Decision Notification: 22 Jun 2022
Greater than low risk review pathway

Dear Mrs Nicolette Soler,

Thank you for submitting an Amendment for the following study;
2020/ETH00132: Development and trial of a sensory assessment measure for children & young people with tic disorders.

The Amendment has been reviewed by the Sydney Children's Hospitals Network Human Research Ethics Committee at its meeting held on 16/06/2022 who have determined the Amendment has been approved.

Notification of an amendment to a research study - General Amendment with form ID 97286

- In the original study protocol we advised that we would provide the study participants with the findings of this study if they choose to receive this. All 16 study participants indicated on their written consent forms that they would want to receive the letter outlining the study results. This amendment application is to seek approval of this letter, outlining the study results, so that it can be provided to the study participants as to thank them and provide them with this information. The results are being provided to study participants, in a letter explaining the findings and a diagram to provide a brief summary of the information provided in the letter.

  As this is a qualitative study, using thematical analysis we also need to do member checking to ensure validity of the study findings as part of the methodology. This requires us to check with study participants that we have interpreted their words and experiences correctly in the study findings. For this reason we have include 10 binary questions (yes/ no responses) for study participants to voluntarily answer to allow them to voice their input and interpretations of the findings prior to publishing themes results. Therefore we are seeking approval of the member checking questionnaire prior to providing this to study participants.

The following documentation is included in this approval:

- 097286_Figure summary of themes and sub themes V1 16.06.2022
- 097286_Notification of an amendment to a research study - General Amendment, 4 June 2022
- 097286_Preliminary results letter to families V1 4.6.2022
- 097286_Questions for member checking V1 04.6.2022

Note to Investigator: The committee noted some typographical and formatting changes were required for the diagram. The following formatting changes were made on behalf of the study team. Please use this document going forward:

- "2.9" was added as the diagram only referenced "2."
- There was a "1)" within the first box, however none of the other boxes had this formatting, and therefore "1)" was deleted to be consistent with the other boxes.
- The font size was increased as the current font size was too small and was difficult to read, especially if printing/mailing the diagram.

It is noted that the Sydney Children's Hospitals Network Human Research Ethics Committee is constituted in accordance with the National Statement on Ethical Conduct in Human Research, 2007 (NHMRC).

This email constitutes ethical and scientific approval only.

For NSW authorised sites (listed in REGIS): A Site General Amendment form will need to be submitted to each affected site. You are not required to upload this form or the ethics approved documents into the site form but you will need to identify this approved amendment form ID (97286).

Should you require any further information, please don't hesitate to contact the Ethics team at SCHN-Ethics@health.nsw.gov.au.

Kind Regards,
Tina
Sent on behalf of Associate Professor Sarah Garnett
Chair, Sydney Children's Hospitals Network Human Research Ethics Committee
<!--[endif]-->
Tina Newman | Research Ethics Officer | Ethics and Governance
e: Tina.Newman@health.nsw.gov.au | w: www.schn.health.nsw.gov.au

<!--[endif]-->Please refer to the SCHN Research Ethics Website for guidance to researcher and sponsors conducting clinical trials and other relevant clinical research impacted by COVID-19: https://www.schn.health.nsw.gov.au/research/ethics-governance/ethics

Cnr Hawkesbury Road and Hainsworth Street, Westmead, NSW Australia
Locked Bag 4001, Westmead 2145, NSW Australia
High Street, Randwick 2031, NSW Australia
%u2672 Please consider the environment before printing this email.

DD /Month /2020

Dear Parent
Parent's Address (To be inserted)

Dear _____,

Re: Invitation for you to participate in a research study

We want to invite you and or your young person to consider your (and / their) participation in a research study titled: **Development and trial of a sensory assessment measure for children & young people with tic disorders.** This research study is conducted by researchers at **The Children's Hospital at Westmead**. It aims to develop a new comprehensive sensory assessment questionnaire for children and young people with tic disorders.

Through the development of this assessment measure, we hope to have a clear understanding of the sensory challenges children and young people with tics may be experiencing. This measure may assist with better care planning with parents and children and young people with tic disorders.

You have been invited to participate because **you meet criteria for one of the following groups:**

a) The **parent(s) / carer(s)** of a child / young person between the ages of 5-16 years old who has a tic disorder and /or

b) A **young person** between the ages of 10-16 years old who has a tic disorder.

Commitment in this study is short term and one-off. You (and or your young person) will be asked to provide a between 30 minutes to 1 hour of your time to this entire study. The interview will currently be conducted via an online meeting using Zoom or over the phone to ensure social distancing and aid with your safety due to the Coronavirus.

A parent (or carer) / young person can participate in this study without the other members of the family participating in the study.

We have attached a participant information sheet (PIS) for parents/ guardians and PIS for young people to this letter which provides more details about this study and describes what participation will involve. If you are a parent of a young person, we kindly ask that you pass on the PIS for young people to your young person too. Knowing what is involved will help you (and / your young person) decide if you (and or they) want to take part in the research. Please read this information carefully.

You (and /your young person) are encouraged to ask questions about anything that you (or they) don't understand or want to know more about.

Participation in this research is voluntary. If you (and / your young person) do not wish to take part, you do not have to. You and your young person will receive the best possible care whether or not you take part.

If you (and / your young person) are interested in participating in this study, we ask that **you complete and sign the consent form attached and email/post it back to the primary research investigator at Nicolette.soler@health.nsw.gov.au**

If we do not hear from you **within 2-4 weeks**, a member of the study team will call you to see if you have received this information and whether you have any questions.

Should you choose to not receive any further contact from the researchers please advise the research team via the phone or email details below or tick the opt-out box on the consent form and return this document to the research team via email / by post using the pre-paid envelope provided.

The contact details for this study are:

Nicolette Soler: (02) 9845-2005 or

email address: nicolette.soler@health.nsw.gov.au or

post: Attention Nicolette Soler, Department of Psychological Medicine, Locked Bag 4001, Westmead, NSW, 2145.

Please do not hesitate to contact us if you require any further information or wish to discuss this study in more detail.

Yours sincerely,

Nicolette Soler
Principal Investigator
Department of Paediatric Neurology& Psychological Medicine,
The Children's Hospital at Westmead

Parent / Guardian Information Sheet

Study Title	Development and trial of a sensory assessment measure for children and young people with tic disorders
Principal Investigator/s & Main Study Contact Person	**Nicolette Soler** **Department of Paediatric Neurology & Psychological Medicine** **The Children's Hospital at Westmead** **(02) 9845-2005 or email address: nicolette.soler@health.nsw.gov.au**

1. Introduction

Thank you for taking the time to read this Information Sheet. This is an invitation for you (and / your young person) to take part in a research study titled: **Development and trial of a sensory assessment measure for children and young people with tic disorders.** The Department of Paediatric Neurology will conduct this study at **The Children's Hospital at Westmead** in conjunction with the Department of Psychological Medicine, The University of Sydney and Health Education and Training Institute (HETI).

This information sheet tells you about the study. It explains the processes involved in taking part. Knowing what is required will help you decide if you (and / your young person) want to take part in this study. Please read this information carefully. Ask questions about anything that you don't understand or want to know more about.

Participation in this research is **voluntary**. If you (and / your young person) do not wish to take part, you do not have to.

2. What is the purpose of this study?

The study aims to understand the experiences of children and young people with tic disorders through:

1) The experiences of parents with a child or young person (5-16 years) with a tic disorder and
2) The experiences of young people (10-16 years) with a tic disorder.

We plan to use this information to create a new sensory-based questionnaire that can be used by doctors and other health professions to **better plan the care of children and young people with tic disorders**. This study allows us to **improve the current questionnaires** that are used commonly.

This research study will be used by the researcher, Nicolette Soler, as part of her research work for her Doctor of Philosophy degree (PhD). This research study is being funded for one year by the Health Education and Training Institute (HETI).

3. Why have I been invited to this study?

You have been invited to participate in this study as you may be:

1) A parent/ career of a child or young person between the ages of 5-16 years with a tic disorder and / or
2) Your young person (10-16 years) with a tic disorder is being invited to participate too.

4. Do I have to take part in this study?

Participation in any research project is voluntary. If you (and / your young person) do not wish to take part, you do not have to. If you (and / your young person) decide that you would like to participate and later change your mind, you (or they) are free to withdraw from the project at any stage.

Your (and / your young person's) decision to not take part, or that you take part and then be withdrawn, will not affect your or your child or young person's routine care, relationship with professional staff or relationship with **The Children's Hospital at Westmead.**

If you do decide that you want to take part, you will be given a consent form to sign, and you will be given a copy to keep. If you are a parent of a young person and your young person chooses to engage in the study, they will need your consent to participate.

5. What does participation in this study involve?

If you (and / your young person) decide that you may take part in this study, we will invite you (and / your young person) to attend **one interview that will run for approximately between 30min to 1 hour** with our researcher. The meeting will be made at a time that is convenient for you (and / your young person). During the period of self-isolation due to the Coronavirus, the interviews will be conducted via Zoom conferencing using a computer or phone device with internet access to assist with ensuring your safety. Interviews may be able to be offered at Children's Hospital at Westmead when deemed appropriate to do so.

If this is not convenient, the interview can be arranged to be conducted over the phone Zoom. The meeting will focus on your child or young person's sensory preferences to assist us with developing a new sensory assessment measure. If the meeting is with your young person, they can choose to have a parent/carer in the interview with them to make them feel more comfortable.

The researcher conducting the interview will ask questions relating to your child/ young person's sensory preferences (i.e. any sensitivity to loud noise, temperature, the feel of clothing items and tags, light lights). The researcher will also ask if any sensory preferences your child / young person is experiencing is impacting on their ability to take part in everyday activities (i.e. dressing, going to school, wearing their school uniform, travel in a car etc.). The researchers have created two pages that list all these sensory experiences and daily life to help prompt any conversation or memories of the sensory experiences your child / young person likes or dislikes.

The interviewer will take notes, and the session will be recorded, with your (and your young person's) consent, to assist with the researchers remembering all the important information that was discussed.

6. What are the possible risks and disadvantages of taking part?

We do not anticipate there to be any risks associated with participating in this study. Should you (and / your young person) feel that any of the questions or discussions are stressful or upsetting, you (or they) can choose not to answer or be involved in the conversations. If you (and / your young person) become upset or distressed as a result of your (or their) participation in the research project, the research team will be able to arrange for counselling or other appropriate support. Any counselling or assistance will be provided by qualified staff who are not members of the research team. This counselling will be provided free of charge. Participants can also contact

Lifeline (telephone number: 13 11 14) at any time or the Kids Helpline (telephone number: 1800 551 800) or Lifeline (telephone number: 13 11 14) at any time.

7. What are the possible benefits of taking part?

We cannot promise that you (and / your young person) will receive any benefits from this research; however, possible benefits may include:

- **By talking about your experiences may give you (and / your young person) a chance to have your (and / their) say about what you (and / they) want health professionals to know about your child/ young persons' (and / their) tic experiences.**
- **Your (and / your young person) involvement may also assist health professionals in asking the correct questions regarding the sensory experiences of children / young people with tics.**

8. Will my child/ young person's medical records be accessed?

If you give permission, the researcher will review your child/ young person's hospital records to confirm details relating to demographics, diagnosis, the severity of tics, presence of other conditions, what medication they are taking and treatment you are receiving. This is to help the researchers obtain accurate information to help with analysis of the research data.

9. What will happen to information about me?

By signing the consent form, you consent to the research team collecting and using the information you provide about your child /young person for this research project. Your privacy and confidentiality, as well as that of your child/ young person, will be protected at all times. Your information will only be used for this research study, and it will only be disclosed with your permission, except as required by law. For example, researchers are required to report if a participant is believed to be at risk of harm.

To protect your privacy, the study team will remove any information that may be used to identify you and your child / your young person from any study documents. Instead of your or your child / young person's name appearing on the documents, study participants will be identified by a specific study code number that applies only to them. Only this code number will be used on any research-related information collected about you and your child / young person for this study so that your and their identity as part of the study will be kept completely private. Only the study team at **The Children's Hospital at Westmead** will have the ability to link this code number with your and your child / young person's personal information, and the linking information will be kept in **a locked filing cabinet in a locked room in the Department of Psychological Medicine**. Your and your child / your young person's data will be stored for **15** of years after the study finishes.

If you (and / your young person) withdraw from the study, we will not collect any more information about you (and/ your young person). We want to keep the information we have already collected about you (and / your young person) to help us ensure that the results of the research project can be adequately measured. Please let us know if you do not want us to do this.

10. How will the results of the study be distributed?

It is anticipated that the results of this research project will be published and/or presented in a variety of forums. In any publication or presentation, the information will be provided in such a way that you and / your young person cannot be identified, except with your explicit permission.

You can indicate on the consent form if you wish to receive a lay summary of the study findings.

11. Who should I contact if I have any questions?

If you (and/ your young person) have any questions or want more information about this study before or during participation, you can contact **Nicolette Soler (Occupational Therapist and study researcher)** on **(02) 9845-2005 /** Nicolette.soler@health.nsw.gov.au.

12. Who do I contact if I have concerns about the study?

All research in Australia involving humans is reviewed by an independent group of people called a Human Research Ethics Committee (HREC). This study has been approved by the Sydney Children's Hospitals Network (SCHN) HREC **(approval number: xxxxxxx).**

If you have any concerns or complaints about any aspect of the project or the way it is being conducted, you may contact the Executive Officer of the SCHN HREC on (02) 9845 1253 or SCHN-Ethics@health.nsw.gov.au.

This Information Sheet is for you to keep. We will also give you a copy of the signed consent form.

Young Person Information Sheet

Study Title	Development and trial of a sensory assessment measure for children and young people with tic disorders
Principal Investigator/s & Main Study Contact Person	**Nicolette Soler** **Department of Paediatric Neurology& Psychological Medicine** **The Children's Hospital at Westmead** **(02) 9845-2005 or email address: nicolette.soler@health.nsw.gov.au**

1. Introduction

You and your parent(s) / carer(s) are invited to participate in a research study titled: **Development and trial of a sensory assessment measure for children and young people with tic disorders.**

The Children's Hospital at Westmead will be conducting this study.

This information sheet tells you about the study. It will help you choose if you want to join or not. You can ask your parent(s), carer(s), friends or doctor, if you need. You do not have to join this study if you do not want to.

Before you think of joining this study, you should know why and how the research is being done. Please read this information sheet carefully.

2. What is the purpose of this study?

The study aims to understand the experiences of children and young people with tic disorders through:

1) Your experiences as a young person with a tic disorder.
2) Your parents /careers experiences if they choose to be part of the study too.

This information will help us to **understand what you are going through**. We plan to use this information to create a new questionnaire that can be used by doctors and other health professions to **better plan the care of children and young people with tic disorders**. This study allows us to **improve the current questionnaires** that are used commonly.

3. Why have I been invited to this study?

You are invited to take part in this study because you are a **young person between the ages of 10-16 with tics.**

4. Do I have to be in this study?

You do not have to participate in this study if you don't want to. The doctors and other health professionals will take the best care of you as they have in the past, regardless of whether you are in the study or not. If you choose to be involved, you can stop being in the study at any time. Just tell one of the researchers or your parent(s)/carer(s) that you don't want to take part anymore.

5. What will happen to me in this study?

If you choose to join the study, you will be invited to attend **one interview that will run for approximately 30 min to 1 hour** at the Children's Hospital at Westmead. You can choose to have your parent(s) / carer(s) in the interview with you. The researcher will ask you questions about your different senses (i.e. smell, taste, touch/ feel, hear, see, movement and balance) and if you have any sensitivities to things (i.e. certain food make you gag, or clothing tags are uncomfortable etc.).

There will be a health professional, Nicolette Soler (you may know as Nicky), who you may already have met before asking you these questions.

Nicky will ask questions to start the discussions happening, make you feel comfortable, answer any questions you may have and take notes. Her job is to understand your experiences with tics. Importantly if you feel sad, upset, or want to talk more about anything that has come up in the chat, she will be there to help you and talk to you afterwards. We can also provide you or your parents/carer with the details of support services to further assist you or provide you with counselling.

Will the researcher read my hospital records?

If you give permission, the researcher (Nicky) will review your hospital records to confirm your age, demographics, diagnosis, the severity of tics, presence of other conditions, what medication you are on and treatment you are receiving. This is to help the research get accurate information to help with the researchers knowing who all the study participants are.

6. Can anything bad happen?

Should you feel that any of the questions or discussions are stressful or upsetting, you can choose not to answer or be involved in the conversations. If anything you talk about during the interview makes you upset, you can stop at any time. Your parent(s)/carer(s) will be given the names of people you can talk to about what is making you upset if that is what you want to do. The researchers can help you do that.

You can also call the Kids Helpline (telephone number: 1800 551 800) or Lifeline (telephone number: 13 11 14) at any time.

7. Will there be any benefits for me in this study?

We cannot promise that you will receive any benefits from this research; however, possible benefits may include:

- **By talking about your experiences may give you a chance to have your say about what you want health professionals to know about you, your experiences and the right questions health professionals need to be asking when they meet you.**

8. How will my privacy be protected?

Your privacy and confidentiality will be protected at all times in this study. This means that unless you allow us, we will not tell anybody else that you are a part of the study or what you said, including your parents/ carers. We will not show any information to anybody else that could be used to identify you unless we are required to do so by law. For example, researchers are required to report if a participant is believed to be at risk of harm.

To protect your privacy, the study team will remove any information that may be used to identify you from any study documents. Instead of your name appearing on them, you will be identified by a specific number that applies only to you. Only this number will be used on any research-related information collected about you for this study. This number allows your identity as part of the study will be kept entirely private. Only the study team at **The Children's Hospital at Westmead** will have the ability to link this code number with your personal information, and the linking information will be kept in **a locked filing cabinet in a locked room in the Department of Psychological Medicine.**

Your data will be stored for 15 years after the study finishes.

If you decide to leave the study, we will not collect any more information about you. We would like to keep the information we have already collected about you to help us ensure that the results of the research project can be measured properly. Please let us or your parent(s) / carer(s) know if you do not want us to do this.

9. What will happen to the study results?

We want to share the study results by publishing them in relevant journal articles and/or presenting them at different conferences. We will make sure that information is published /presented in such a way that you are not identifiable unless you and your parent(s) /carer(s) have permitted us to do so.

You / your parent(s) or carer(s) can also tell us on the consent form if you want to receive a simple summary of the study findings for information.

10. Who should I contact if I have any questions?

If you have any questions or want more information about this study before or during participation, you can talk to **Nicolette Soler** (Nicky) **(Occupational Therapist and study researcher)** on **(02) 9845-2005 / Nicolette.soler@health.nsw.gov.au**.

You can also ask your parent(s)/carer(s) to talk to us.

11. Who do I contact if I have concerns about the study?

All research in Australia involving humans is reviewed by an independent group of people called a Human Research Ethics Committee (HREC). This study has been approved by the Sydney Children's Hospitals Network HREC **(approval number: xxxxxxx)**.

Please talk to your parent(s)/carer(s) if you are worried about being in this study, or you have a complaint. They can speak with **Nicolette Soler** on **(02) 9845-2005,** or they can contact the Human Research Ethics Committee on (02) 9845 1253 or SCHN-Ethics@health.nsw.gov.au.

Parent / Guardian & Young Person Consent Form

Study Title	Development and trial of a sensory assessment measure for children and young people with tic disorders
Principal Investigator & Main Study Contact Person	**Nicolette Soler** **Department of Paediatric Neurology& Psychological Medicine** **The Children's Hospital at Westmead** **(02) 9845-2005 or email address: nicolette.soler@health.nsw.gov.au**

Declaration by Parent / Guardian

☐ I have read the Parent / Guardian Information Sheet and the Young Person Information Sheet or someone has read it to me in a language that I understand.

☐ I understand the purposes, procedures and risks of the research project described in the Parent / Guardian Information Sheet.

☐ I have had the opportunity to ask questions, and I am satisfied with the answers I have received.

☐ I freely agree to my young person (children between the ages of 10-16 years) participating in this research project as described and understand that I am free to withdraw myself or my young person at any time during the project without affecting my or their future health care.

☐ I understand that I will be given a signed copy of this document to keep.

☐ I consent to the interview being conducted with myself and /my young person to be recorded. I understand that the recording is only to be used to aid the researchers with transcribing the interview and will then be destroyed.

☐ I permit the study researcher to review my child / young person's medical records at The Children's Hospital at Westmead to gain information concerning my child / young person's condition and treatment for this project. I understand that such information will remain private and confidential.

☐ I wish to receive a lay summary of the study findings via the following email/post address:

_____

Opt-out from any further information from study:

☐ I choose not to receive any further information about this study. I understand that following the researchers being made aware that I do not want to be further contacted I will not be provided with any further information about this study.

_____

Name of Young Person (please print): _____

Signature of Young Person: _____ Date: _____

Name of Parent / Guardian (please print): _____

Signature of Parent / Guardian: _____ Date: _____

Under certain circumstances (see Note for Guidance on Good Clinical Practice CPMP/ICH/135/95 at 4.8.9) a witness to informed consent is required.*

Guide for responding to study participants' potential to psychological distress in the study:
Development and trial of a sensory assessment measure for children & young people with tic disorders.

The well-being of participants is of utmost importance. This research will be conducted to the highest ethical standard and per the NHMRC National Statement on Ethical Conduct in Human Research and approved by all relevant Ethics Committees.

All appropriate care will be given to participants and their families. All individuals will be informed that choosing not to take part in the study, or withdrawing from the study at any stage, will not adversely affect their or their child or young person's care in any way. It is possible that thinking about the health of oneself or one's family member may elicit distress in some participants. This situation will be considered case-by-case.
To address this possibility, the research team will:

1. Ensure that participants have an understanding of what the research entails prior to consent.
2. Explicitly state the option of withdrawal and termination at any time due to distress.
3. Offer support options to any participants who express distress or a desire for support. Participants will be referred to appropriate counselling services, as needed, including referral to the treating centre's psychologist or social worker for further assessment. All Participant Information Sheets will also provide the details for research team contacts should distress arise after study completion.

As is standard practice, any adverse events will be formally documented and reported to the appropriate HREC(s) within 48 hours.

Follow-Up Procedures for Participants Experiencing Distress

- The safety of the participant is of the highest priority. Participants experiencing distress may contact the research team after the interview. All contact points and outcomes will be documented in the study database.
- If the participant is over 16 years of age, a member of the research team will contact the participant within 5 working days to discuss how they are feeling and to offer a referral for support.
- If the participant is under 16 years of age, a member of the research team will contact his or her parent/carer to discuss the participant's distress and appropriate services that might be helpful. The research officer will also ask to speak with the participant who is under 16 years to offer them a referral for support.
- If a participant does not answer their phone, a voice message will be left with contact details for the research team. Up to 10 attempts will be made to contact participants, with a maximum of 2 attempts made per day (i.e., one in the morning and one in the afternoon). No more than two messages will ever be left for participants who are not in contact with the Research Team and require follow-up. If phone contact cannot be made, the research team will send a sensitively-worded email detailing support services (see below for a list of services)
- If phone contact is made, an example script (see page 5) can be used to help guide the conversation with the participant.
- After the name of a support service (or health professional) has been provided to the participant (either by phone contact, email or letter), a member of the Research Team can attempt to contact the participant again if there are any ongoing concerns or to support the participant with engagement with the referral service, **if appropriate and clinically indicated**.

Script for speaking with the participant

“Hi, this is [YOUR NAME] from the development and trial of a sensory assessment measure for children & young people with tic disorders study. Thank you very much for participating in the study. I’m calling today to follow-up with you and wonder if this is a good time to talk?’’

(If no, schedule an appropriate time to call back and document in the database).

(If yes) ‘’Thank you. I am aware that you identified feelings of distress after completing (Choose: the semi-structured interviews or the online questionnaires). Is it OK if I ask you a few questions about how you are feeling?

- Are you still feeling that way?
- How long have you been feeling like this?
- It is not uncommon for people to feel this way, particularly when they have been under stress.
- Have you spoken with anyone about how you have been feeling or sought any help? ‘’

(If yes) ‘’Are you still connected with these support services? Have they been helpful for you?’’

(If no) ‘’There are support services available. Would you be interested in connecting with support?’’

‘’Are you in regular contact with your GP, psychologist, or other health providers (as appropriate to participant)?’’

Direct participant to the most appropriate support service or referral pathway using the list below. There may also be additional services unique to the organisation you are working in.

‘’Thank you for speaking with me today. If you need any other information on how to access any support or assistance for yourself (or your child/ young person), please feel free to call me on 9845 2005.’’

POTENTIAL REFERRAL SERVICES

- **General Practitioner**
  Can provide individuals with a Mental Health Care Plan, which allows them to see a psychologist for at least 10 sessions a year. There are two streams for psychology services, which patients can access through Medicare with a referral from their GP:

  Better Access to Mental Health Care Scheme
  http://www.health.gov.au/internet/main/publishing.nsf/content/mental-ba-fact-pat

  Access to Allied Psychological Services (ATAPS)
  http://www.health.gov.au/internet/main/publishing.nsf/content/mental-boimhc-ataps

- **Australian Psychological Society – Find a Psychologist Service**
  www.psychology.org.au/FindaPsychologist
  This online service allows you to search for psychologists in a particular area, who are in private practice and provide services for a fee.

- **beyondblue**
  www.beyondblue.org.au
  This is a national, independent, not for profit organisation working to address issues associated with depression, anxiety and related substance misuse disorders.
  beyondblue has a 24-hour line: **1300 22 4636**.
  This is not a free call, it will cost the same as a local call from a landline but might be more from a mobile.

- **Black Dog Institute**
  www.blackdoginstitute.org.au
  This is a not for profit organisation offering expertise in depression and bipolar disorder.

- **Lifeline**
  www.lifeline.org.au
  This is a national charity providing all Australians experiencing a personal crisis with access to 24-hour crisis support and suicide prevention services.
  Lifeline has a 24-hour crisis line: **13 11 14**.

- **Kids Helpline**
  www.kidshelp.com.au
  This is a not-for-profit organisation offering a counselling service for Australian children and young people aged between 5 and 25 years.
  Kids Helpline: **1800 55 1800**

Appendix II: : Supplementary documents from research studies (Chapters 3 to 6)

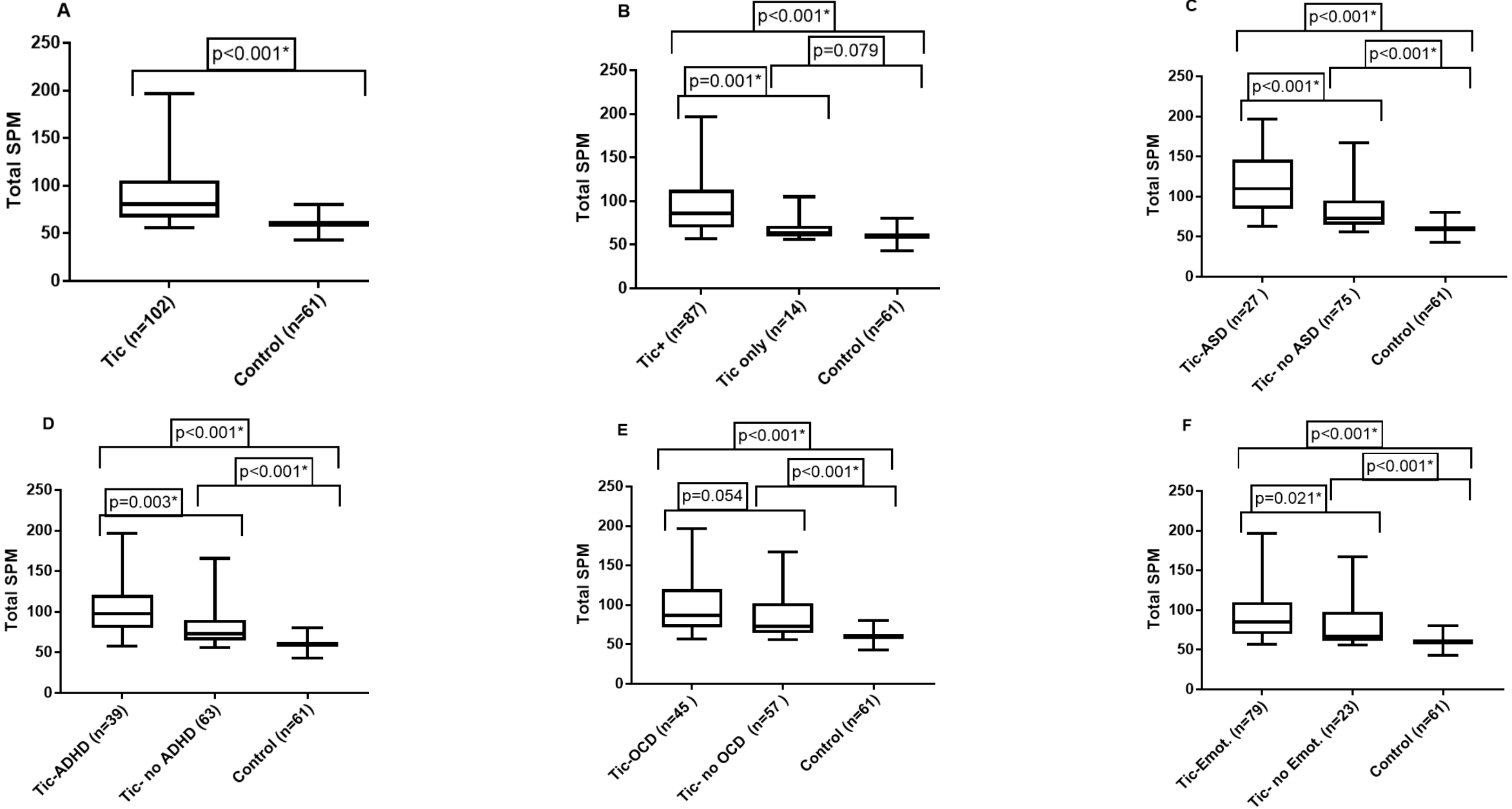


Figure S1: Sensory dysregulation associated with tic disorders and comorbidity.

Sensory Processing Measure (SPM) results show elevated sensory dysregulation in tic participants compared to controls (S1A). Sensory dysregulation were not elevated in tics only group (S1B). The presence of all comorbidities, ASD (S1C), ADHD (S1D), OCD (S1E) and emotional disorder (S1F) was associated with increased sensory scores.

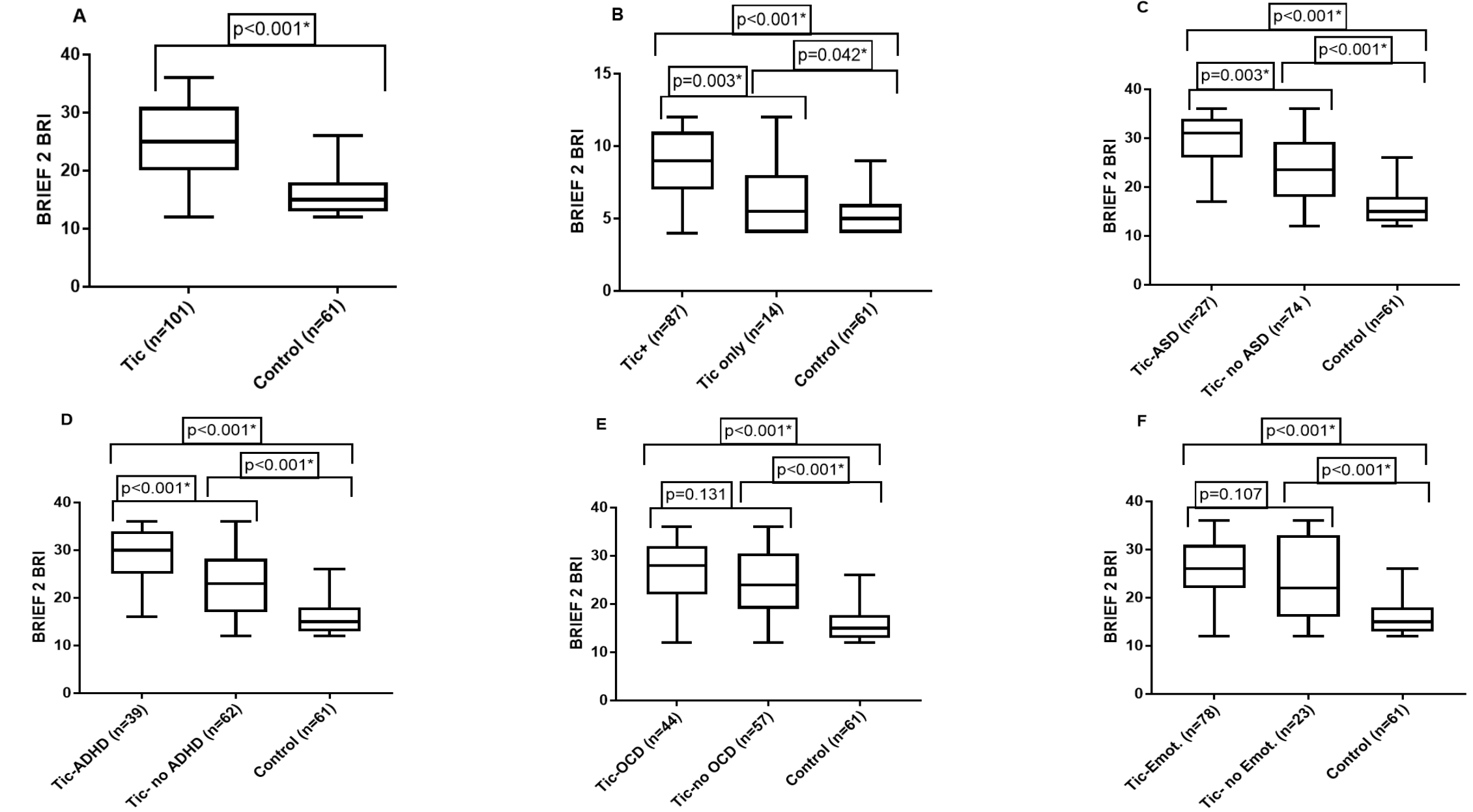


Figure S2:Executive dysfunction (Behaviour)associated with tic disorders and comorbidity.

The Behavior Rating Index (BRI) on the Behavior Rating Index of Executive Function-2 (BRIEF-2) shows elevated executive function difficulties in tics participants compared to controls (S2A); Executive function difficulties were not present in the tics only participants (S2B). The presence of all comorbidities, ASD (S2C), ADHD (2SD), OCD (S2E) and emotional disorder (S2F), was associated with higher executive function difficulties.

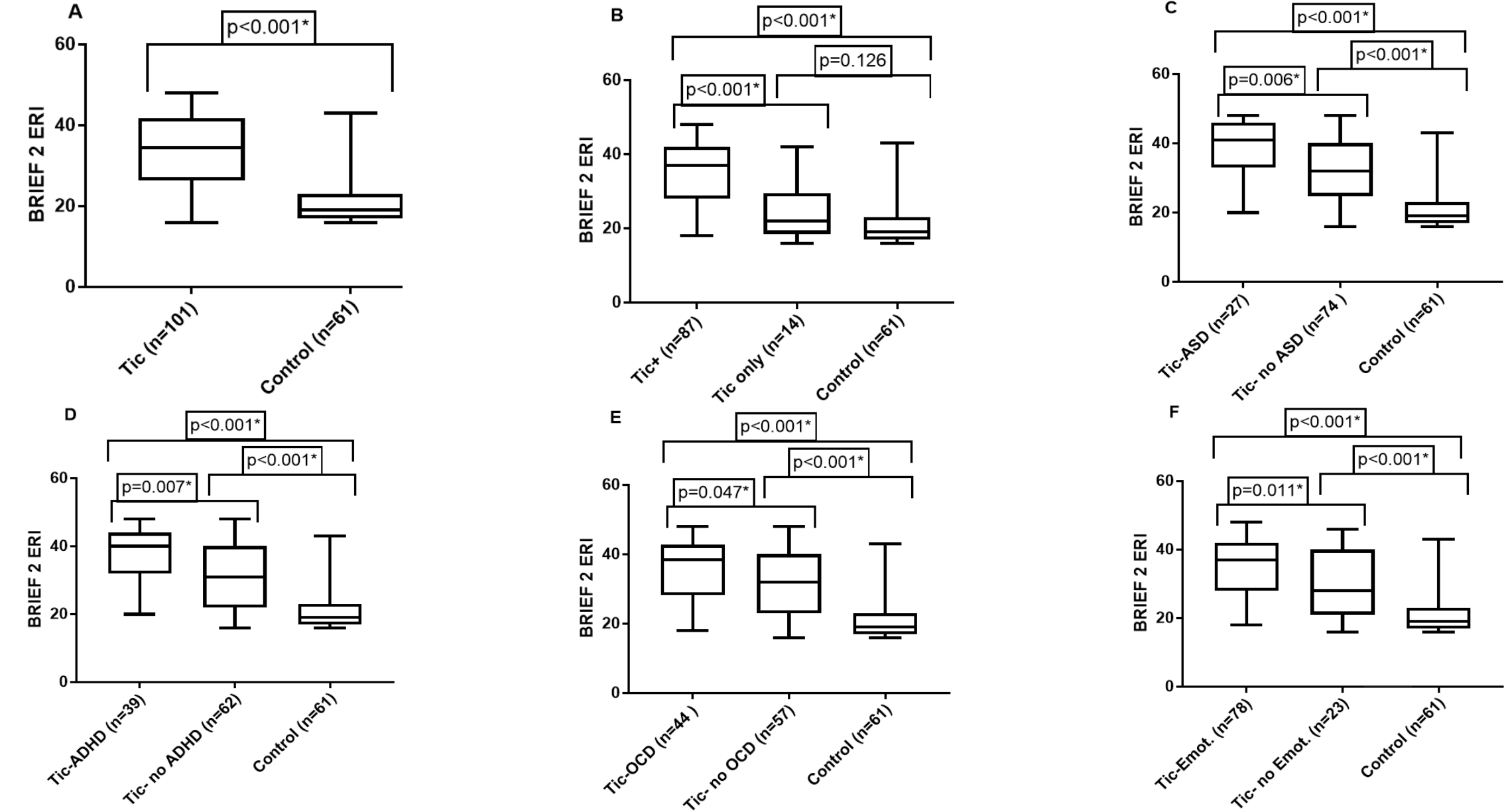


Figure S3: Executive dysfunction (Emotional dysfunction) associated with tic disorders and comorbidity.

The Emotional Rating Index (ERI) on the Behavior Rating Index of Executive Function-2 (BRIEF-2) shows elevated executive function difficulties in tics participants compared to controls (S3A); Executive function difficulties were not present in the tics only participants (S3B). The presence of all comorbidities, ASD (S3C), ADHD (S3D), OCD (S3E) and emotional disorder (S3F), was associated with higher executive function difficulties.

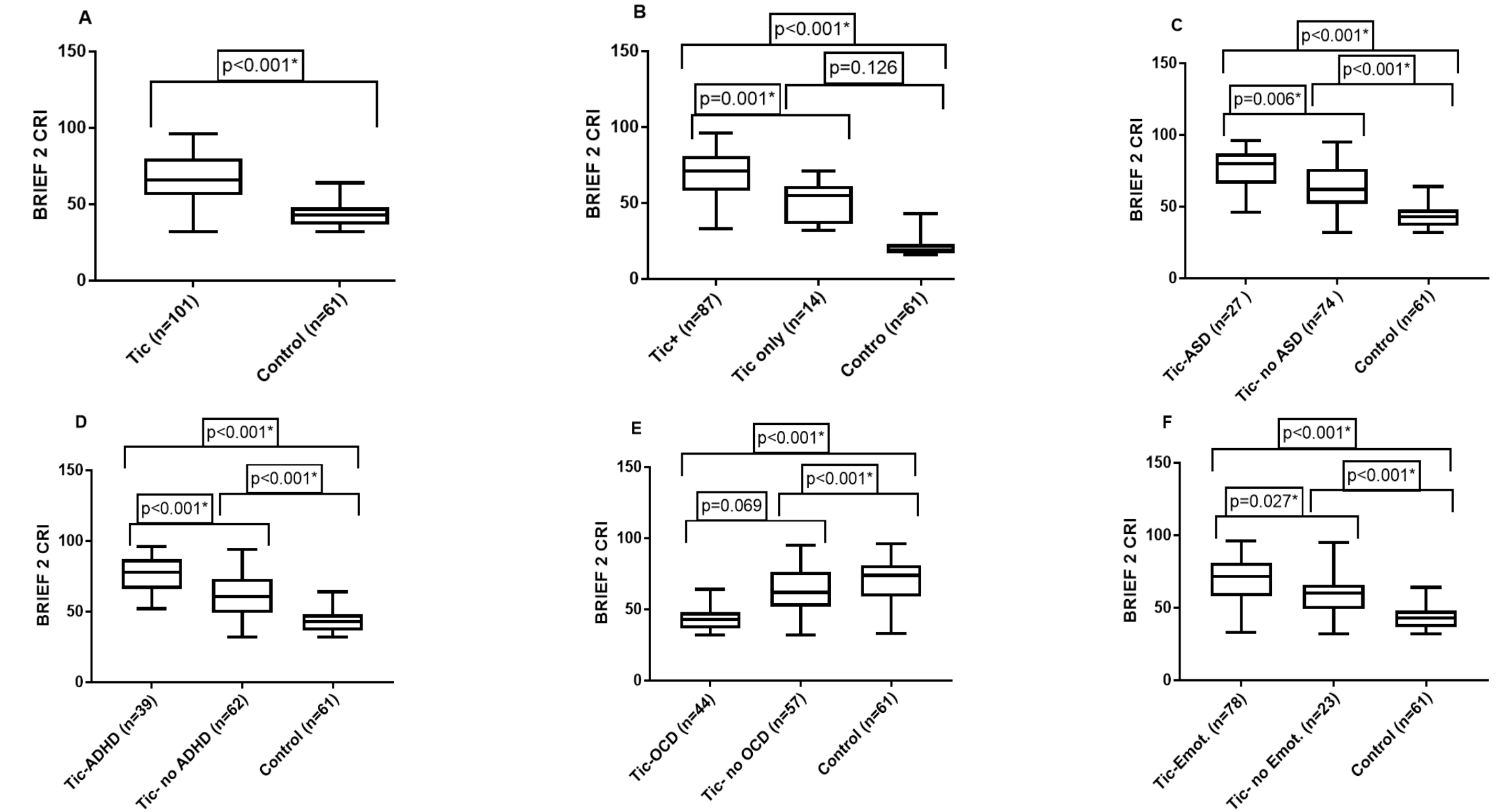


Figure S4: Executive dysfunction (Cognitive dysfunction) associated with tic disorders and comorbidity.

The Cognitive Rating Index (CRI) on the Behavior Rating Index of Executive Function-2 (BRIEF-2) shows elevated executive function difficulties in tics participants compared to controls (S4A); Executive function difficulties were not present in the tics only participants (S4B). The presence of all comorbidities, ASD (S4C), ADHD (S4D), OCD (S4E) and emotional disorder (S4F), was associated with higher executive function difficulties.

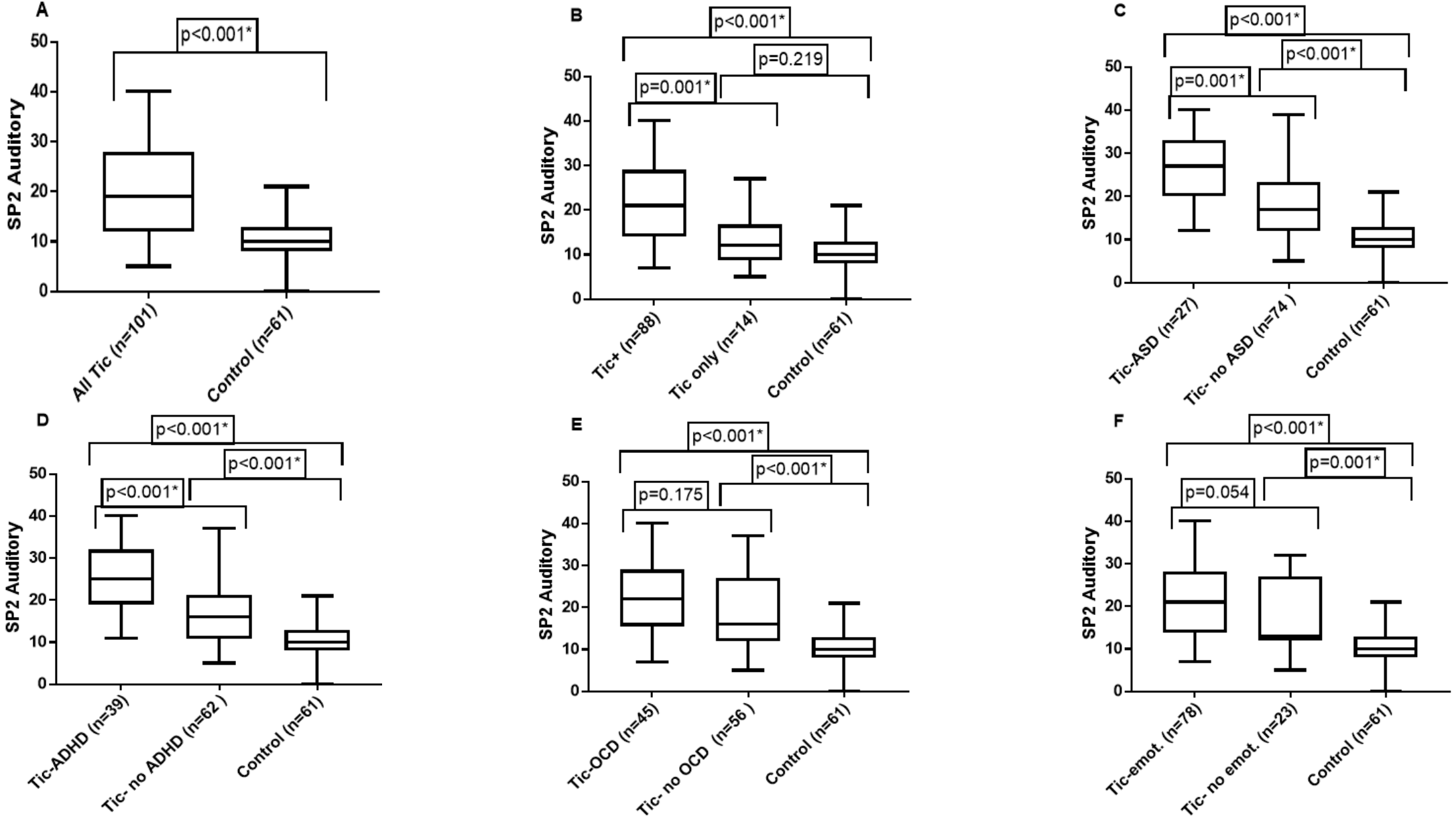


Figure S5: Auditory sensory dysregulation associated with tic disorders and comorbidity.

Auditory sensory dysregulation (sub-scale) on the Child Sensory Profile 2 (SP2) is elevated in tics participants compared to controls (S5A); and is more prevalence in children with tic and comorbidities than in children with tics only (S5B). The presence of all comorbidities, ASD (S5C), ADHD (S5D), OCD (S5E) and emotional disorder (S5F), was associated with higher auditory sensory dysregulation.

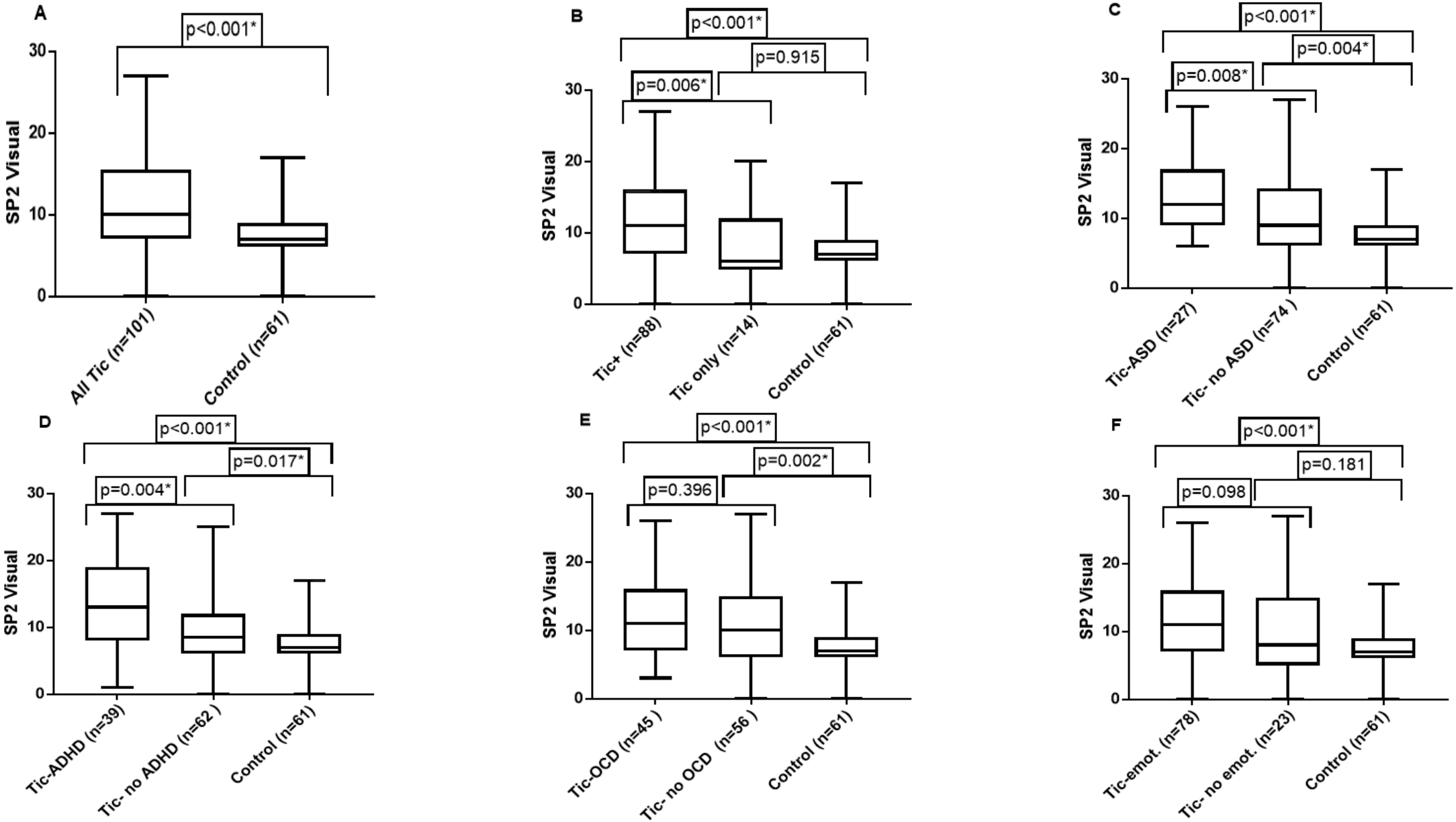


Figure S6: Visual sensory dysregulation associated with tic disorders and comorbidity.

Visual sensory dysregulation (sub-scale) on the Child Sensory Profile 2 (SP2) is elevated in tics participants compared to controls (S6A); and is more prevalence in children with tic and comorbidities than in children with tics only (S6B). The presence of all comorbidities, ASD (S6C), ADHD (S6D), OCD (S6E) and emotional disorder (S6F), was associated with higher visual sensory dysregulation.

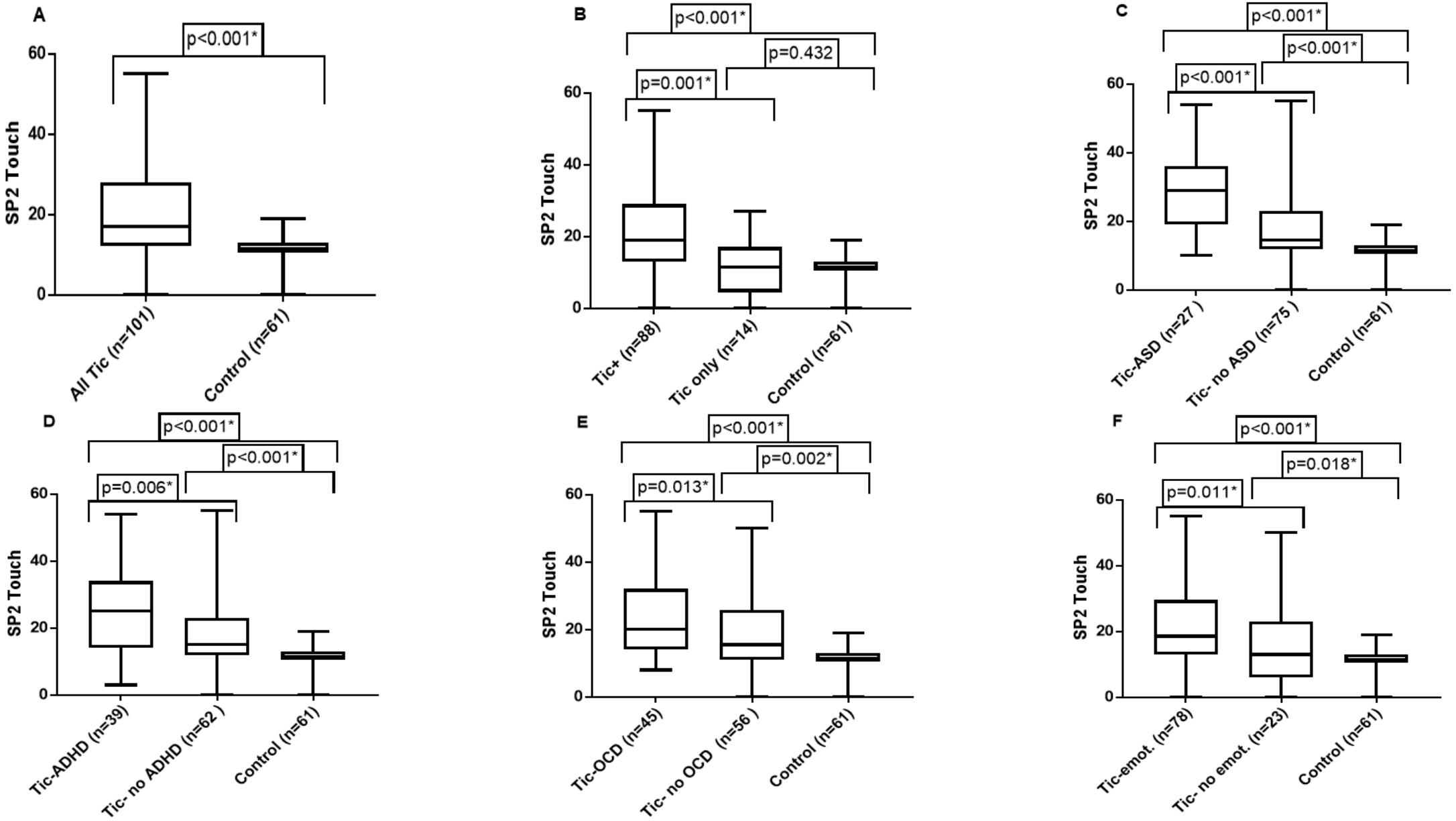


Figure S7: Tactile sensory dysregulation associated with tic disorders and comorbidity.

Tactile (touch) sensory dysregulation (sub-scale) on the Child Sensory Profile 2 (SP2) is elevated in tics participants compared to controls (S5A); and is more prevalence in children with tic and comorbidities than in children with tics only (S7B). The presence of all comorbidities, ASD (S7C), ADHD (S7D), OCD (S7E) and emotional disorder (S7F), was associated with higher tactile sensory dysregulation.

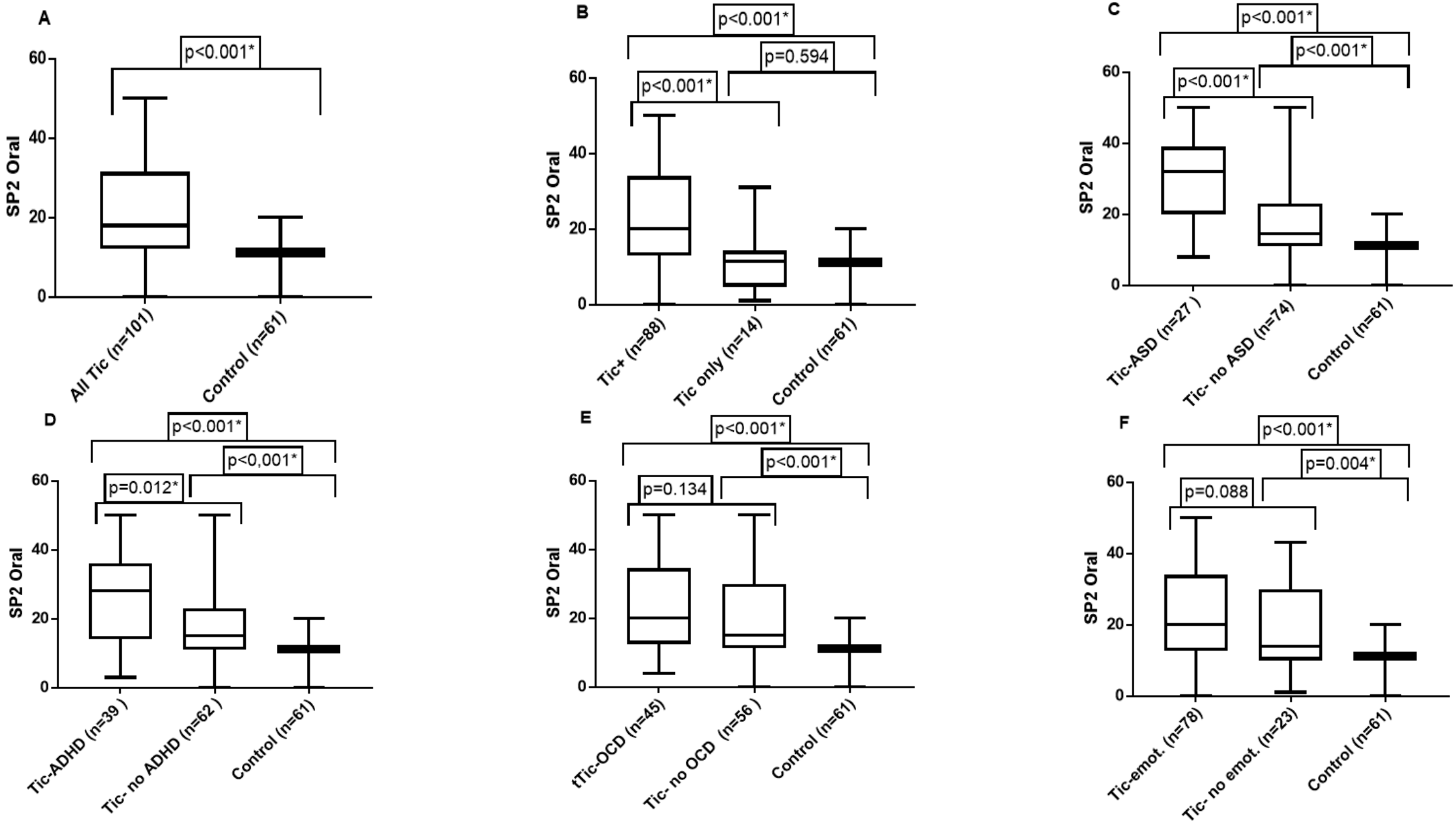


Figure S8: Oral (taste and smell) sensory dysregulation associated with tic disorders and comorbidity.

Oral sensory dysregulation (sub-scale) on the Child Sensory Profile 2 (SP2) is elevated in tics participants compared to controls (S8A); and is more prevalence in children with tic and comorbidities than in children with tics only (S8B). The presence of all comorbidities, ASD (S8C), ADHD (S8D), OCD (S8E) and emotional disorder (S8F), was associated with higher oral sensory dysregulation.

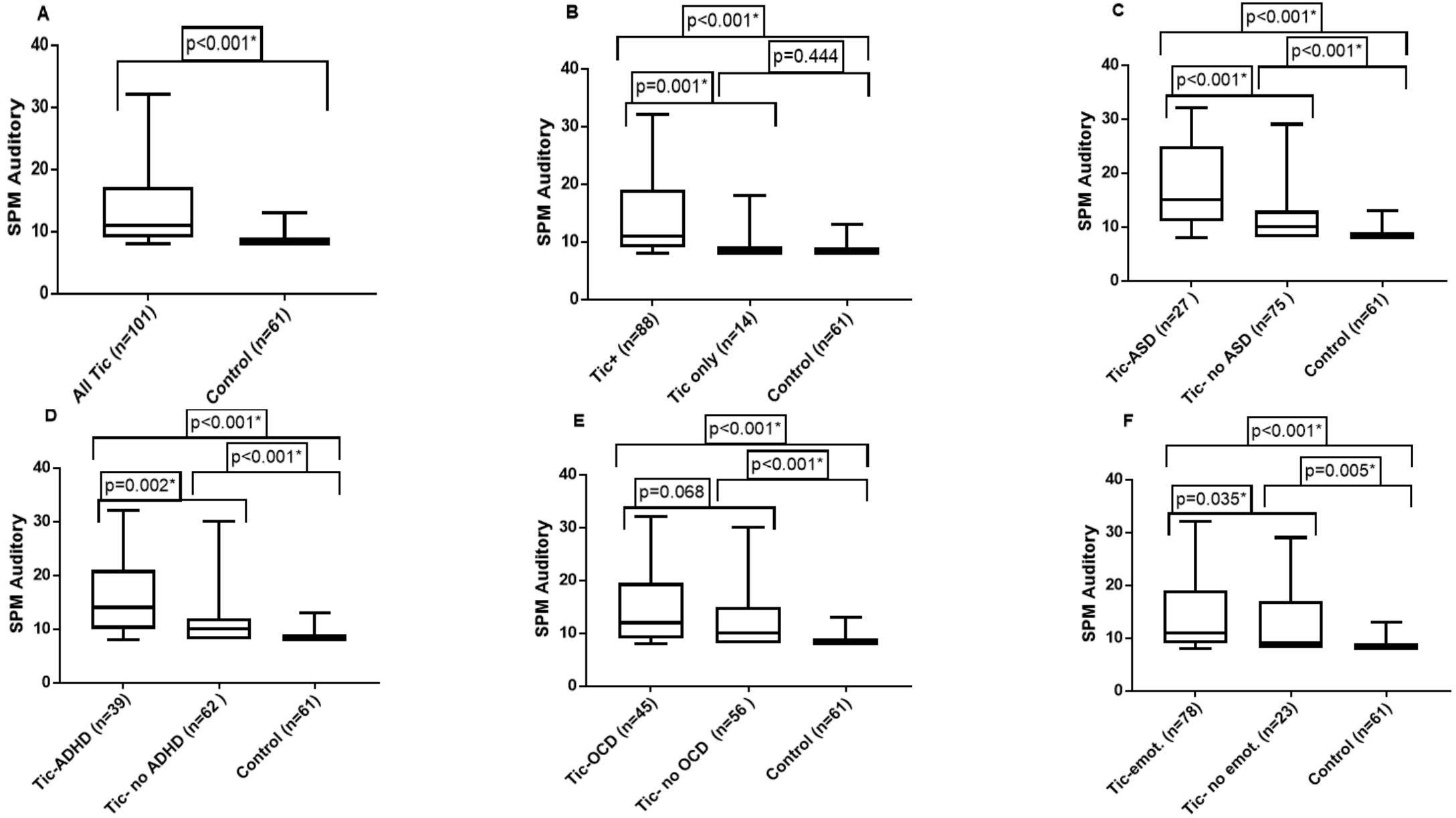


Figure S9: Auditory sensory dysregulation associated with tic disorders and comorbidity.

Auditory sensory dysregulation (sub-scale) on the Sensory Processing Measure (SPM) is elevated in tics participants compared to controls (S9A); and is more prevalence in children with tic and comorbidities than in children with tics only (S9B). The presence of all comorbidities, ASD (S9C), ADHD (S9D), OCD (S9E) and emotional disorder (S9F), was associated with higher auditory sensory dysregulation.

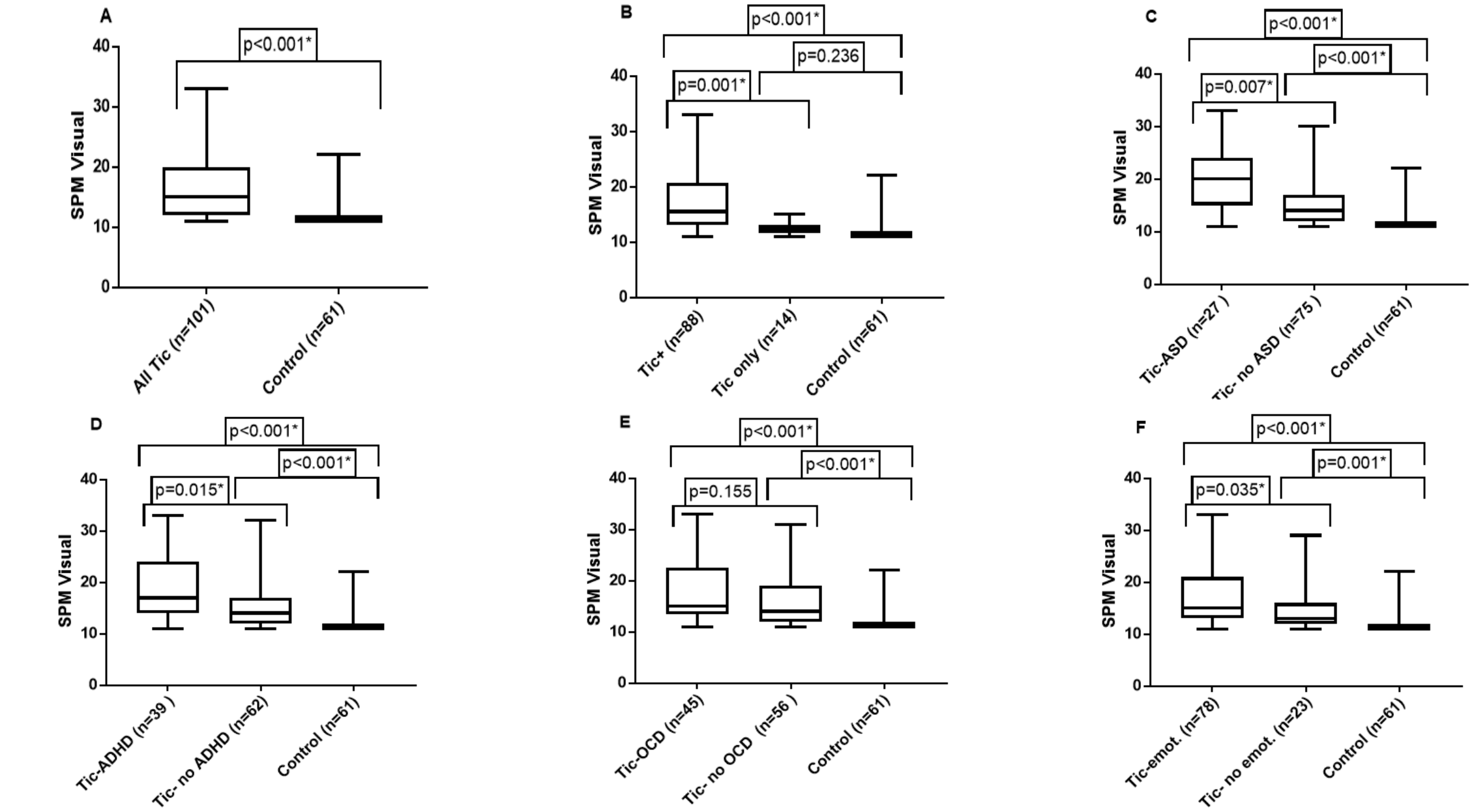


Figure S10: Visual sensory dysregulation associated with tic disorders and comorbidity.

Visual sensory dysregulation (sub-scale) on the Sensory Processing Measure (SPM) is elevated in tics participants compared to controls (S10A); and is more prevalence in children with tic and comorbidities than in children with tics only (S10B). The presence of all comorbidities, ASD (S10C), ADHD (S10D), OCD (S10E) and emotional disorder (S10F), was associated with higher visual sensory dysregulation.

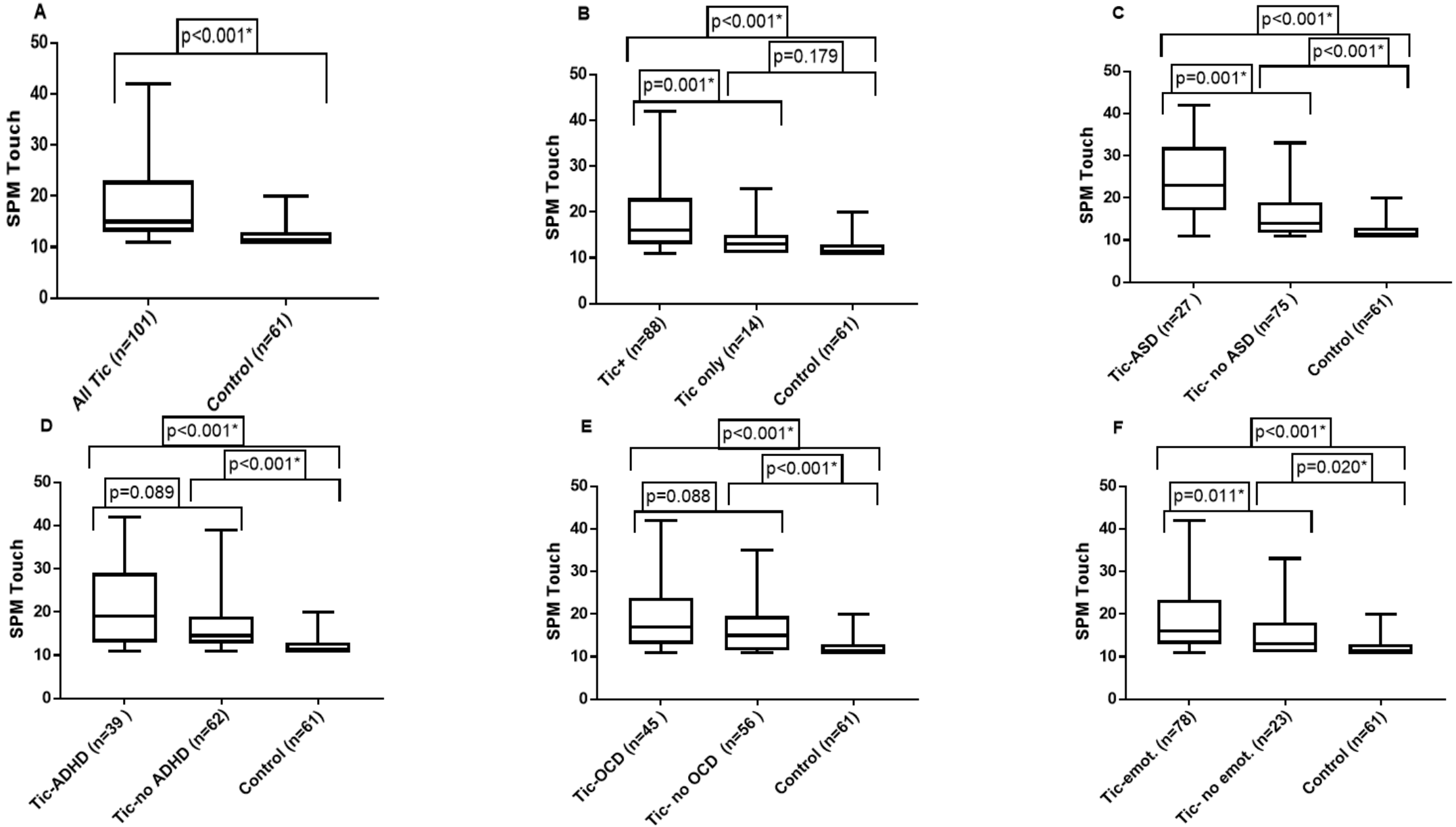


Figure S11: Tactile (Touch) sensory dysregulation associated with tic disorders and comorbidity.

Tactile sensory dysregulation (sub-scale) on the Sensory Processing Measure (SPM) is elevated in tics participants compared to controls (S11A); and is more prevalence in children with tic and comorbidities than in children with tics only (S11B). The presence of all comorbidities, ASD (S11C), ADHD (S11D), OCD (S11E) and emotional disorder (S11F), was associated with higher tactile sensory dysregulation.

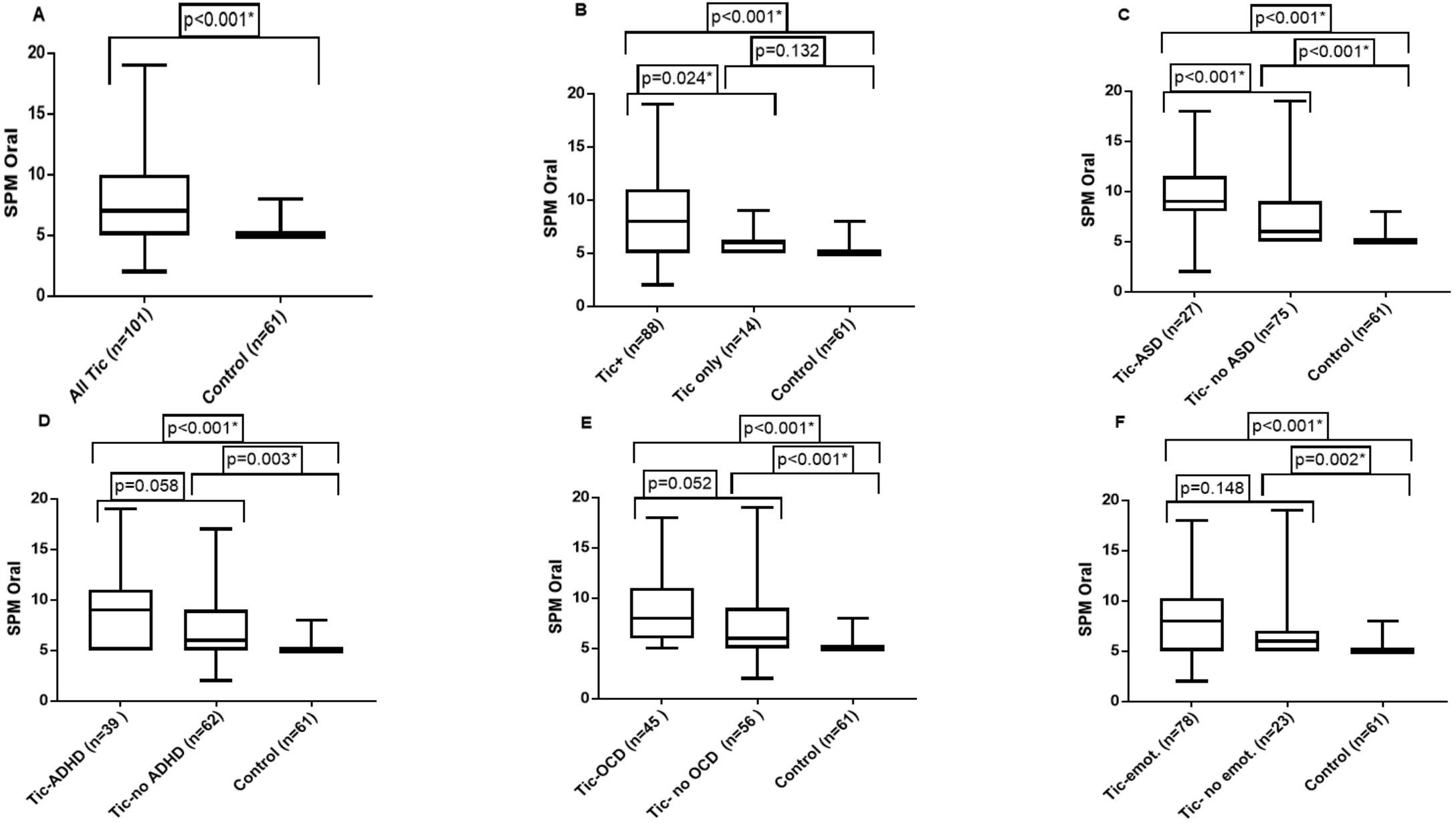


Figure S12: Oral (Taste and Smell) sensory dysregulation associated with tic disorders and comorbidity.

Oral sensory dysregulation (sub-scale) on the Sensory Processing Measure (SPM) is elevated in tics participants compared to controls (S12A); and is more prevalence in children with tic and comorbidities than in children with tics only (S12B). The presence of all comorbidities, ASD (s12C), ADHD (S12D), OCD (S12E) and emotional disorder (S12F), was associated with higher oral sensory dysregulation.

Appendix S1: PRISMA-P Protocol

Prisma- P Protocol

Administrative information

1a. Title

An evaluation of sensory assessment tools for children or adolescents with neurodevelopmental disorders: protocol for a systematic review.

To determine the most appropriate sensory tools for children or adolescents with neurodevelopmental disorders are available for clinicians to use with their patients?

2) What are the psychometric properties of the sensory assessment tools for children or adolescents with neurodevelopmental disorders?

3) Are the sensory assessment tools for children or adolescents with neurodevelopmental disorders sensitive and specific enough to use with a particular neurodevelopmental disorders, such as Tourette's syndrome?

4) Is there a need for a new sensory assessment tools for children or adolescents with neurodevelopmental disorders?

1b.Update

This systematic review protocol is not an update of a previous systematic review.

2. Registration

This systematic review protocol was registered with the International Prospective Register of Systematic Reviews (PROSPERO), as per the PRISMA-P guidelines, on the 4th of December 2019.

3a. Authors contact details

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3b. Contributions

Mrs Nicolette Soler is the guarantor of this systematic review.

All authors have read and approved the final version of the manuscript, with all contributors acknowledged. All authors contributed towards the conception and organization of the research project. Dr. Paula Bray, Prof. Russell Dale and Nicolette Soler contributed to the development and refinement of the research question, the draft protocol, development of the selection criteria, risk of bias assessment strategy, data extraction criteria and provided critical revision. Nicolette Soler developed the search strategy with assistance from Dr Paula Bray and librarians, Tess Aitken, (academic liaison librarian with University of Sydney) and Trish Bennett (library manager at the Children's Hospital at Westmead). Dr Paula Bray provided statistical expertise. Dr Iain Perkes and Chris Hardwick critically reviewed and approved the final version of the protocol.

4. Amendment

Nil amendments proposed at present in relation to this protocol.

Should amendments to this protocol be required, the description of each proposed change including the rationale for the change and date will be tracked through amendments made to the amendments section of protocol and the changes will be registered with Prospero.

5. Support

5a. Financial and other support for review

This systematic review is not funded specifically or by an institution but the following authors are receipts of the following funding grant through their research positions and employment contracts:

- Mrs N. Soler is the recipient of a Petre Foundation Fellowship.
- Professor R.C. Dale is the recipient of a Petre Foundation Fellowship and a National Health and Medical Research Council (NHRMC) Practitioner Fellowship funded by the Australian Government.
- Dr I.E. Perkes is the recipient of a NHMRC Medical Postgraduate Scholarship (RG162061, 2017-2020).

5b. Review funder and / or sponsor

Although the authors are the recipients of fellowship grants, there is no funder for this systematic review and no funding has been received to conduct this study. The authors are affiliated with The Children's Hospital at Westmead and the University of Sydney and therefore these two institutions assume overall responsibility for this systematic review and control over the data.

5c. Role of sponsor or funder

This systematic review is being conducted independently by staff employed through The Children's Hospital at Westmead and the University of Sydney, Children's Hospital Westmead Clinical School, Sydney, Australia as part of the primary author's PhD topic. Therefore there are no sponsors or review funder for this review.

Introduction

6. Rational

Tic disorders, although understood to be a neurodevelopmental movement disorder due to the repetitive, stereotypical, rapid, non-rhythmic movements or vocalizations can be conceptualised as a sensorimotor phenomenon (1, 2, 3). When vocal and motor tics are present for more than a 12 month period, the condition is referred to as Tourette's syndrome. Most patients, regardless if they have a tic disorder or Tourette's syndrome, report not only a need to obtain a 'just right' feeling in their body but also a premonitory urge, which is a sensory discomfort which precedes the tic and it is believed may be involved in tic expression (3,4). There have been broader sensory dysregulation symptoms described both structurally and functionally in people with tic disorders. Deficits in filtering irrelevant sensory stimuli, known as sensory motor gating, has been identified in individuals with tic disorders (5, 6). Structural sensorimotor abnormities have been identified in this same cohort through brain imaging studies including: electrophysiological studies (6, 7), magnetoencephalography (8) functional imaging (9) and volumetric imaging (10).

Sensory dysregulation results in disproportional behavioural responses to the sensory experiences due to the impaired ability to manage internal or external sensory input (11, 12). These sensitivities to sensory input can result in dysfunction in five main areas, namely impaired i) social skills; ii) adaptive responses; iii) self-esteem; iv) daily living skills; v) gross-, fine- and sensory-motor development (13). There are many terms used to describe sensory symptoms in neurodevelopmental disorders including: atypical sensory reactivity, sensory phenomena, sensory processing and sensory modulation. In this protocol, the term sensory dysregulation will be used to describe sensory impairment in children with tics (7, 11, 14, 15).

Tic disorders are neurodevelopmental conditions as tics begin in childhood and typically coexist with other neurodevelopmental and neuropsychiatric conditions in 80-90% of cases (16). Sensory dsyregulation symptoms are common, and amplified in the presence of these neurodevelopmental and neuropsychiatric conditions including autism spectrum disorder, obsessive compulsive disorder, attention deficit hyperactivity disorder and depressive disorders (including anxiety) (17, 18).

Through our recent study investigating the prevalence of sensory symptoms in children with tic disorders and comorbidities it was identified that the sensory assessment tools had limitations (19). Children with tics or their parents anecdotally identified additional functional sensory impairments not captured by the measures (the Sensory Profile 2 (20) and the Sensory Processing Measure (21)). Difficulties such as intolerance to human made noises, such as chewing, sucking or cutlery scraping on crockery was not recorded on these commonly used measures. This results in clinicians not being able to measure the true significance of the impairment thus goal setting and treatment planning is problematic. A systematic review is warranted to understand if any other valid and reliable sensory measurement tools exist and are appropriate for use in this population.

Thus far a systematic review has been conducted evaluating the sensory measures used in infants for the first two years of life (22) but the same review has not been conducted in children older than two years of

age. As tics are predominantly diagnosed in childhood and are the most common movement disorder in children, it is imperative that we have appropriate and reliable assessment measures to use with this population (1, 2, 19).

7. Objectives

The focus of this systematic review is evaluation of the quality (psychometric properties) of all tools used to measure sensory dysregulation in children or adolescents with tic disorders or other neurodevelopmental disorders and comorbidities as listed in the Diagnostic and Statistical Manual 5 (DSM-5) (23). For this review the term neurodevelopmental disorders includes but not limited to: autism spectrum disorder; Asperger's; attention-deficit/ hyperactivity disorder; obsessive compulsive disorder; communication disorder; intellectual disability; learning disorders; global developmental delay; language disorders and tic disorders.

The construct of interest is 'sensory dysregulation', the population is children or adolescents with a neurodevelopmental disorder, the type of instruments of interest are all sensory measurement tools and 'all' measurement properties are explored in the review.

We will determine if existing tools are reliable, valid, specific, sensitive and responsive to assess the complex sensory dysregulation difficulties that patients with tics or associated comorbidities experience. Furthermore, we will establish if there is a need to develop a new sensory measure which is more sensitive in identifying these patients' functional deficits.

Methods

8. Eligibility Criteria

Types of Study to be included

We will include all types of studies but case studies will be summarised separately. Case reports, case series or any study with n<5 will be excluded and unpublished research will not be considered. Studies that describe cohorts of mixed age (i.e. youth (12-24 years)) will only be include if there is subgroup analysis provided or if we are able to request data from authors where no subgroup analysis is provided. Studies that involve mixed diagnostic groups (e.g., Tourette syndrome and obsessive compulsive disorder) will be included.

Planned Population

Research studies which report on individual participants or the participant group age range of between 3 years through to 18 years of age with a neurodevelopmental disorder will be included. Age expressed in either months or years by the study will be acceptable as long as the study recruited children or adolescents between 3 and 18 years of age. Should the group participants' mean age and age ranges not be reported on in the study the reviews will assess the age criteria of the assessment tools used in the study. If the eligible age range for the assessment tool used in the study is between 3 years and 18 years of age, then studies will be included in the review. We will include studies that report on adults and children / adolescents whereby the child and or adolescent data is reported separately from the data relating to adult participants. Participants in the study younger than 3 years of age (as they would be classified as infants or toddlers rather than children) or older than 18 years would be ineligible.

We based this diagnostic list on the NDD section of the DSM 5. However, because diagnosis is not a central interest to this study, we included any diagnostic method so long as we could approximate a diagnostic classification

For the purposes of this review we define Neurodevelopmental Disorders (NDD) as any of the following:

- autism spectrum disorder (any severity, including DSM IV equivalent diagnosis i.e. Asperger's) ;
- attention deficit hyperactivity disorder (any type including ICD equivalent e.g. hyperkinetic disorder);
- anxiety (any, including social anxiety disorder, separation anxiety disorder);
- depressive disorder;
- tic disorders (including vocal / motor tic disorder or Tourette's syndrome etc.) and
- obsessive compulsive disorder diagnosed through the Diagnostic and Statistical Manual 5 (DSM-5) (23) criteria, will be included.

Because the method of diagnosis is not a construct of interest, this review will accept any method of diagnostic ascertainment if the diagnosis is reported. We expect that we may not be able to record the diagnostic method in terms of agent (self-report, trained interviewer, or clinician) and technique (symptom scale, unstructured, semi-structured, or structured interview; self-report forms) extrapolated to diagnosis or diagnosis made by another health professional or expert (i.e. the referring agent; the child/ adolescents' paediatrician, GP or health care expert etc.) as this information may not be provided. Should this be the case, meta-analysis will not be able to be conducted on agent, form and method of diagnosis unless this information can be accessed by researchers. Individuals with any condition other than a neurodevelopmental condition already listed will be excluded from the systematic review.

Studies which identify children or adolescents to be non-verbal or experience profound intellectual disability will be included if the study used proxy reported questionnaires (i.e. parent rated outcome measures) or adapted the assessment measures to allow for the participants to provide adequate and accurate means for self-report.

Intervention

Any sensory assessment measure used to assess sensory dysregulation in a paediatric population with a neurodevelopmental disorder will be included in the review. Any form of sensory related screening tool or assessment measure or questionnaire or instrument that is parent, child/ adolescent/ teacher or clinician rated will be included. All sensory assessment measures used to assess sensory sensitivities in children or adolescents with neurodevelopmental disorders will be evaluated using COSMIN (24, 25, 26). The article needs to report on psychometric properties of the sensory measurement tool of interest.

Articles which do not report the psychometric properties of the various sensory assessment measures or tools will be excluded. All studies involving therapy or intervention or treatment approaches or strategies for neurodevelopmental conditions or sensory symptoms will be excluded, even if these studies used sensory measures as outcome tools. Should findings from other assessment tools other than sensory measures be reported in the study, these findings will be excluded.

Comparator(s) / control

No comparators or controls.

Outcomes

Each included sensory measure will be assessed according to COSMIN (Consensus-based Standards for the selection of health status Measurement Instruments) criteria: validity (discriminant ability and convergent validity), reliability (intra- and inter-rater), responsiveness, sensitivity and specificity of the measure or tool.

Timing
There will be no date restriction in relation to the publication or development of the sensory assessment tools reported on in the articles. Assessment tools that are no longer available for clinicians to use as they are surpassed or out-dated will be summarised separately.

Setting
Participants can be of any cultural or ethnic background and the study could have been completed in any country and there are no restrictions to setting type (e.g., hospital or community).

The researchers will exclude any unpublished materials or abstracts from the search.

Language
We will include articles reported in English and in other languages where a copy of the translated original article can be obtained easily in English. If relevant titles are found in other languages, the authors will list these in the supplementary documentation.

9. Information Sources
We will search the following electronic bibliographic databases: Medline, Medline in Process, PsycINFO, EMBASE, AMED through Ovid, Scopus, CINAHL, Web of Science, Informit, Cochrane Library, Complementary Medicine and 'Google Scholar'. We will hand search the contents lists of relevant journals and articles found.

In addition to searching databases, the following clinical trial registries will be reviewed to obtain information on evidence that may soon be available: Australian New Zealand Clinical Trials Registry; ClinicalTrials.gov; The European Union Clinical Trials Register; Systematic Reviews and Cochrane.

10.Search Strategy
The search strategy was developed in conjunction with researchers and librarians from both The Children's Hospital at Westmead and The University of Sydney with expertise in systemic review search strategies. The preliminary Medline (via Ovid) search strategy was developed first. Appendix 1 below provides a final version of the Medline (via Ovid) search strategy. This final version of search terms was adapted to each of the following databases: Medline in Process, PsycINFO, EMBASE, AMED through Ovid, Scopus, CINAHL, Web of Science, Informit, Cochrane Library, Complementary Medicine and 'Google Scholar'.

This search will include only quantitative studies unless any qualitative studies report on the psychometric properties of a sensory assessment measure as a mixed methods study. The search strategy will not impose any limits in relation to dates, setting or study designs. Due to resource restrictions, articles in a language other than English, will be excluded from the review.

The search strategy will include only terms relating to or describing the psychometric properties of screening or assessment of tools, questionnaires or measures or instruments or surveys relating to sensory symptoms in children and adolescents with neurodevelopmental disorders.

Approaches to building the search strategies include:

- Broad searches are to be conducted to ensure the breadth of sensory assessment measures in children with a neurodevelopmental disorder are included.
- A combination of subject heading and keyword searching will be used where available (Ovid platforms, CINAHL).

- Across databases, identical search strategies are to be utilised where possible.
- Search terms identified for the core search will include tic disorders and /comorbid associated neurodevelopmental disorders as well as, Gilles de la Tourette Syndrome; Tourette's or tic disorder; chronic tic disorder; provisional tic disorder; pervasive tic disorder; vocal tic or motor tics; ASD; autism spectrum disorder; attention-deficit/ hyperactivity disorder; obsessive compulsive disorder; OCD; communication disorder; intellectual disability; learning disorders; global developmental delay; language disorders further will not be added (Informit, Cochrane Library).
- The total number of citations retrieved for the core search will be exported for coding.

To ensure that there is no duplication of this systematic review, PROSPERO has been searched and will continue to be reviewed during the length of this study.

11. Study Records

11a. Data management

The citations identified through the searches will be imported into EndNote for management of the review process and any duplicate copies of studies will be removed. An excel spread sheet will be used by the reviewers to assess and track eligibility of the remaining articles as this program allows for collaboration amongst reviewers. All reviewers will be trained in the use of these software programs prior to the commencement of this review.

11b. Selection process

A two-pass selection process will be used to identify relevant articles. This will be conducted in duplicate by two independent reviewers (PB and NS).

First Pass: In the first pass two reviewers will independently assess all studies identified via the database search against the clinical question and eligibility criteria based on information contained in the title, abstract and description (including MESH headings) to identify studies that potentially meet the inclusion or exclusion criteria outlined above. Any duplicate copies of studies will be removed and number of papers will be recorded. Studies identified for inclusion in the first pass by the duplicate reviewers will be compared. If there is disagreement between reviewers, an additional independent reviewer (RD) will be consulted to enable consensus to be reached. Where eligibility is unclear, the study will be reviewed at second pass.

Second Pass: Full articles of studies included in the first pass will be obtained and assessed against the clinical question and eligibility criteria as well as the COSMIN Risk of Bias checklist (24, 25, 26). Studies identified for inclusion in the second pass by the duplicate reviewers will be compared. If there is disagreement between reviewers, an additional independent reviewer will be consulted to enable consensus to be reached. Studies remaining after the second pass will go forward to data extraction and evidence grading. Following second pass, the reference lists of all included papers will be searched for any publications that have not been identified in the database search. Full papers will be retrieved and reviewed for eligibility.

A record will be kept outlining the reason for each articles exclusion. Neither of the reviewers (PB and NS) will be blinded to any information pertaining to the study articles under review, this including the title, abstract, authors or intuitions.

11c. Data collection process

Two independent reviewers will carry out the evaluation and data extraction independently (PB and NS). The two data extractions will be compared. Where there is disagreement between reviewers, an additional reviewer (RD) from the current research group will be consulted.

Excel programs will be used to store, track and evaluate the extracted data from the included studies for assessment of study quality and evidence synthesis.

12.Data items

We will follow a two-step process to the review as outlined by the COSMIN Risk of Bias checklist (24, 25,26), i) evaluate the quality of the studies (risk of bias) and the quality of the measurement tool (measurement properties) and ii) then use the evidence to decision framework to inform the recommendations.

Extracted information will be based on the COSMIN Risk of Bias checklist for systematic reviews of patient reported outcome measure 2017 (24, 25, 26) and will include: patient reported outcome measure development, content validity, structural validity, internal consistency, cross-cultural validity\measurement invariance, reliability, measurement error, criterion validity, hypotheses testing for construct validity, responsiveness, sample size, age range, mean age, standard deviation of age, sex distribution, Cronbach's alpha, reliability coefficients, time interval between administration of measures, Kappa coefficient, and minimal important change or difference.

In addition, the following data will be extracted: diagnosis; comorbid diagnoses; diagnostic method; severity of neurodevelopmental disorder symptoms, study design; study location; study setting; study population; participant demographics; baseline characteristics; sensory assessment measures used; primary purpose of assessment tool; other purposes; age range of assessment tool; type of test; normative sample; domains tested; components tested; time to administer; method of measurement use (e.g. informant or self-report); manual/ equipment required; training required; scoring; interpretation of scores; cost of assessment; test-retest; intra- and inter-rater reliability; discriminant ability; assess evidence of mis-fitting items / persons; show good overall model fit; study duration; other relevant outcomes assessed; adverse events from assessment and affiliation and source of funds.

13.Outcomes and prioritization

Primary Outcomes

The primary outcome of this review will be to identify the quantity and quality of sensory measures used in children or adolescents with tics disorders or neurodevelopmental conditions. Following the identification of these tools, the psychometric properties will be evaluated using COSMIN Risk of Bias checklist for systematic reviews (24, 25, 26) through a two-step processes, the evaluation of the study criteria and the risk of bias assessment. Following the pooling of data at the end of these processes, the reviewers aim to establish the efficacy of the tools assessed and provide recommendations through the use an evidence based decision framework.

Secondary outcome:

Through the extraction of data pertaining to each sensory measurement tool, it would be understood which individual assessment components or questions on the various tools are most application, valid and reliable over and above the psychometric properties of the whole measure if this information is

provided to the authors. Should there be no valid or reliable to that best fits the needs of children or adolescents with tic disorders or other neurodevelopmental disorders, the information gathered on the individual assessment components of the different tools may assist with understanding of the framework for the development of a new sensory measure.

14. Risk of bias and individual studies

Two review authors (PB & NS) will independently assess the risk of bias using the COSMIN Risk of Bias checklist (24, 25, 26). Reviewer (PB) has previous experience of conducting multiple systematic reviews and brings to the study her expertise with previous risk of bias experience. The following domains will be evaluated: study design and sampling; sample size; representativeness of the sample; use of comparison / normative sample group; incomplete outcome data; selective reporting. We will grade these domains as a low, unclear or high risk of bias. Any disagreement between reviewers over risk of bias in of the studies will be resolved by discussion and a third reviewer (RD) will be consulted if necessary. Although reviewers will independently screen and assess, reviewers will not be blinded to the studies.

15. Data synthesis

15a. Criteria for quantitative data synthesis

As this review is assessing the psychometric properties of assessment measures rather than an intervention, it is not foreseen that it would be appropriate or feasible for analysis of meta-analyses of the data.

15b. Describe planned summary methods, data handling & methods of combining data from data for appropriate quantitative studies

Following a table of evidence being generated for the clinical question, the reviewers will use Rasch Analysis, a unique mathematical modelling based approach allowing the measurement of persons and items on a the same scale which has equal-interval property of the scale. Rasch modelling facilitates analysis of responsiveness of individual items on the measurement tool with respect to their calibrated positions within a measure. The use of the Rasch measurement model will provide robust analysis of the internal construct validity of sensory dysregulation outcome measures used in children or adolescents with tic disorders or associated neurodevelopmental or neuropsychiatric conditions. The measures will be evaluated in regards to the even spread of item values; precision and reduced error of measurement; probability and improbability (fit) of item and person values to that expected from the model; overall reliability; simplicity and conformity to the nature of the clinical values that are being measured.

The reviewers will attempt to contact the original authors to obtain relevant missing data should this occur. Should the desired data not be reported on, the reviewers re aware that such data may need to be reconstructed from other statics such as p values. Extracted data will be analysed using SPSS (version 25) software.

15c. Any additional proposed analyses

If the necessary data is available, subgroup analysis of the effectiveness of the various measures in relation to specific neurodevelopmental or neuropsychiatric groups will be analysed. This will allow the reviewers to understand if the various tools identified can be used effectively across different diagnosis under the umbrella terms of neurodevelopmental or neuropsychiatric conditions or if the tools can only be appropriately used with certain specified conditions.

Additional information, such as the cost of the various tools identified; the training required for clinicians to administer the tools; timing to administer the tools; duration before a test-retest tool can be re-administered would be additional information extracted and analysed.

15d. Type of summary planned if quantitative synthesis is not appropriate

Should meta-analysis not be appropriate, the reviewers will include qualitative synthesis of the data in the form of text and tables to summaries the findings and data extracted.

16. Meta-bias(es)

It is expected by the reviewers that a publication bias will be difficult to assess in this review due to a lack of registries for studies relating to measurement properties and development of measurement tools. Therefore the reviewers are aware that it may not be possible to take this factor into account in the methodology of this review. Where possible and available the reviewers will assess for and report on the risk of publication bias. The reviewers will search for any published study protocols and compare these to published papers on the findings of the study if available, to determine if any significant differences appear or data reported on. Should this information be available, the reviewers will ensure to assess that the findings reported in the articles under the results section are the same findings reported on in the discussion and conclusion of the paper.

17. Confidence in cumulative evidence

COSMIN Risk of Bias was designed to assess and evaluate the properties of assessment tool in relation to the methodological quality of the study. The quality of the evidence will be evaluated using the COSMIN Risk of Bias checklist (24, 25, 26) to assess the strength of the body of evidence relating to the psychometric properties of the sensory assessment tools The quality of the evidence will be evaluated as high, moderate, low or very low as defined in table five of the COSMIN manual for Systematic reviews of PROMS (24, 25, 26).

Appendix 1: Subject index terms for Medline (via OVID)

Population:

(Age)

1. adolescen*.mp. or Adolescent/
2. child*.mp. or exp Child/
3. p*ediatric
4. preschool*.mp.
5. school age.mp.
6. teen*
7. youth*
8. 1 OR 2 OR 3 OR 4 OR 5 OR 6 OR 7

(Diagnosis)

9. neurodevelopmental disorders/ or neurodevelopment* disorder*.mp.
10. developmental disorders/
11. neuropsychiatric disorder*
12. anxiety.mp.
13. depressive disorder.mp.
14. attention deficit disorder.mp.
15. autism spectrum.mp. or autism Spectrum Disorder/
16. exp Communication Disorders/ or communication disorder*.mp.

17. Intellectual Disability/ or intellectual* disab*.mp.
18. Learning Disorders/
19. global developmental delay*.mp.obsessive compulsive disorder.mp.
20. intellectual development* disorder*.mp.
21. exp Language Disorders/
22. obsessive compulsive disorder*.mp.
23. exp Tic Disorders/ or tic disorder*.mp.
24. tourette*.mp.
25. 9 OR 10 OR 11 OR 12 OR 13 OR 14 OR 15 OR 16 OR 17 OR 18 OR 19 OR 20 OR 21 OR 22 OR 24 OR 24
26. **8 AND 25**

Intervention:

27. atypical sensory reactivity.mp.
28. (process* adj1 (sensory or sense*)).mp.
29. sensorimotor.mp.
30. sensory dysregulation.mp.
31. sensory defensiveness.mp.
32. sensory discrimination.mp.
33. ((sensory or sense*) adj1 (overrespons* or over-respons*)).mp.
34. somatic hypersensitivity.mp.
35. sensory modulation.mp.
36. sensory over* reactivity.mp.
37. sensory perception.mp.
38. sensory reactivity.mp.
39. sensory seeking.mp.
40. somatosensory.mp
41. 27 OR 28 OR 29 OR 30 OR 31 OR 32 OR 33 OR 34 OR 35 OR 36 OR 37 OR 38 OR 39 OR 40

42. assess*.mp.
43. evaluat*.mp.
44. checklist*.mp. or Checklist/
45. instrument*.mp.
46. instrumentation.mp.
47. inventor*.mp.
48. measure*.mp.
49. profile*.mp.
50. screen*.mp.
51. survey*.mp.
52. tool*.mp.
53. test*.mp.
54. questionnaire*.mp.
55. survey*.mp.
56. validation Stud*.mp.
57. 42 OR 43 OR 44 OR 45 OR 46 OR 47 OR 48 OR 49 OR 50 OR 51 OR 52 OR 53 OR 54 OR 55 OR 56
58. **41 AND 57**

Outcome:

(Search terms were developed and adapted through recommendations from published article By C.B. Terwee 2009) (27)

59. Alpha.mp.
60. factor Analysis, Statistical/
61. ceiling effect.mp.

62. coefficient*.mp.
63. coefficient of variation.mp.
64. computer adaptive testing.mp.
65. Concordance*.mp.
66. cronbach*.mp.
67. cross-cultural equivalence.mp.
68. dimensionality*.mp.
69. differential item functioning.mp.
70. DIF.mp.
71. “discriminant analysis”/
72. discriminative.mp.
73. factor*analys*s
74. factor structure*.mp.
75. findings.tw.
76. floor effect.mp.
77. generali?a*.tw.
78. individual variability.mp.
79. int?r*rater*. mp.
80. int?r*tester*
81. intra*examiner*.mp.
82. inter*assay*.mp.
83. interval variability.mp.
84. intra*assay*.mp.
85. intra-assay*.mp.
86. intra*individual*.mp.
87. intra*participant*.mp.
88. inter*observer*.mp.
89. inter*technician*.mp.
90. intra*observer.mp.
91. intra*technician.mp.
92. inter*examiner*.mp.
93. interscale correlation*.mp.
94. inter*individual*.mp.
95. inter*participant*.mp.
96. “internal consistency”/
97. IRT.mp.
98. item bank.mp.
99. “item correlation”/
100. item discriminant.mp.
101. “item selection”/
102. “item reduction”/
103. item response model.mp.
104. kappa*.mp
105. meaningful change*.mp.
106. minimal detectable concentration.mp.
107. minimal*detectable*change*.mp.
108. minimal*detectable*difference*.mp.
109. minimal* important change.mp.
110. minimal* important difference.mp.
111. minimal*real*difference*.mp.
112. minimal* real*change.mp.
113. multitrait*scaling*analysis*.mp.
114. outcome*.mp.
115. “observer variation*.mp.
116. precision.tw.

117. psychometric*.mp. or Psychometrics/
118. precision.tw.
119. precise values.tw.
120. quotient*.mp.
121. rasch.mp.
122. rate variability.mp.
123. reliability.MP
124. responsiv*.mp.
125. re-test*.mp.
126. retest.mp.
127. result*.tw.
128. repeatab*.tw
129. (repeated.tw.) AND (measure*.tw)
130. replicab*.tw.
131. reproducib*.mp.
132. ''reproducibility of results''/
133. sensitiv*.mp.
134. sensitivity.mp.
135. stability.mp.
136. standard error of measurement.mp.
137. specificity.mp.
138. "Sensitivity and Specificity"/
139. subscale*mp.
140. valid*.mp.
141. variability analysis.mp.
142. 59 OR 60 OR 61 OR 62 OR 63 OR 64 OR 65 OR 66 OR 67 OR 68 OR 69 OR 70 OR 71 OR 72 OR 73 OR 74 OR 75 OR 76 OR 77 OR 78 OR 79 OR 80 OR 81 OR 82 OR 83 OR 84 OR 85 OR 86 OR 87 OR 88 OR 89 OR 90 OR 91 OR 92 OR 93 OR 94 OR 95 OR 96 OR 97 OR 98 OR 99 OR 100 OR 101 OR 102 OR 103 OR 104 OR 105 OR 106 OR 107 OR 108 OR 109 OR 110 OR 111 OR 112 OR 112 OR 113 OR 114 OR 115 OR 116 OR 117 OR 118 OR 119 OR 120 OR 121 OR 122 OR 123 OR 124 OR 125 OR 126 OR 127 OR 128 OR 129 OR 130 OR 131 OR 132 OR 133 OR 134 OR 135 OR 136 OR 137 OR 138 OR 139 OR 140 OR 141
143. 26 and 58 and 142

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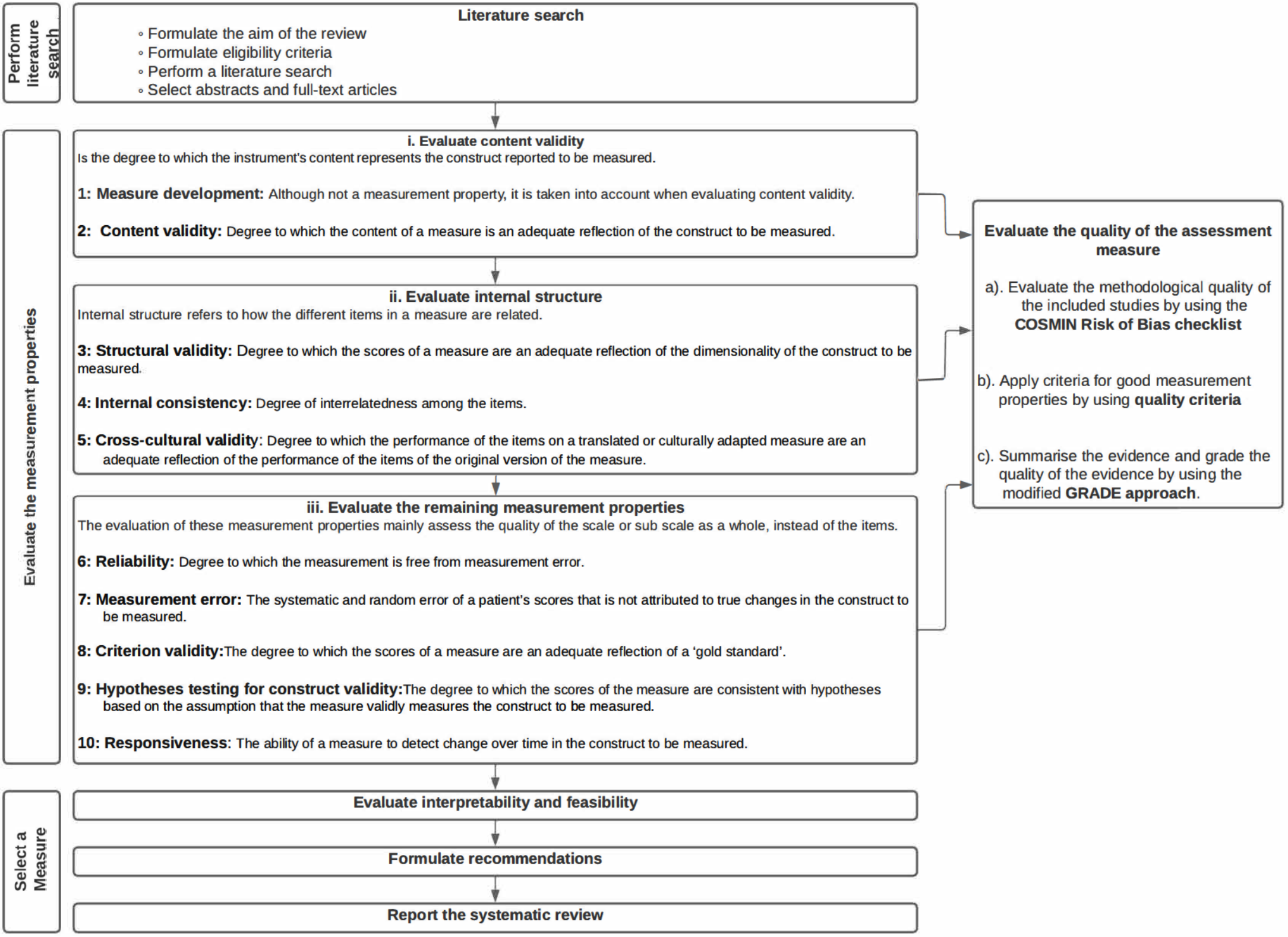

Perform literature search
Literature search
◦ Formulate the aim of the review
◦ Formulate eligibility criteria
◦ Perform a literature search
◦ Select abstracts and full-text articles
Evaluate the measurement properties
i. Evaluate content validity
Is the degree to which the instrument's content represents the construct reported to be measured.
1: Measure development: Although not a measurement property, it is taken into account when evaluating content validity.
2: Content validity: Degree to which the content of a measure is an adequate reflection of the construct to be measured.
ii. Evaluate internal structure
Internal structure refers to how the different items in a measure are related.
3: Structural validity: Degree to which the scores of a measure are an adequate reflection of the dimensionality of the construct to be measured.
4: Internal consistency: Degree of interrelatedness among the items.
5: Cross-cultural validity: Degree to which the performance of the items on a translated or culturally adapted measure are an adequate reflection of the performance of the items of the original version of the measure.
iii. Evaluate the remaining measurement properties
The evaluation of these measurement properties mainly assess the quality of the scale or sub scale as a whole, instead of the items.
6: Reliability: Degree to which the measurement is free from measurement error.
7: Measurement error: The systematic and random error of a patient's scores that is not attributed to true changes in the construct to be measured.
8: Criterion validity: The degree to which the scores of a measure are an adequate reflection of a 'gold standard'.
9: Hypotheses testing for construct validity: The degree to which the scores of the measure are consistent with hypotheses based on the assumption that the measure validly measures the construct to be measured.
10: Responsiveness: The ability of a measure to detect change over time in the construct to be measured.
Evaluate the quality of the assessment measure
a). Evaluate the methodological quality of the included studies by using the COSMIN Risk of Bias checklist
b). Apply criteria for good measurement properties by using quality criteria
c). Summarise the evidence and grade the quality of the evidence by using the modified GRADE approach.
Select a Measure
Evaluate interpretability and feasibility
Formulate recommendations
Report the systematic review

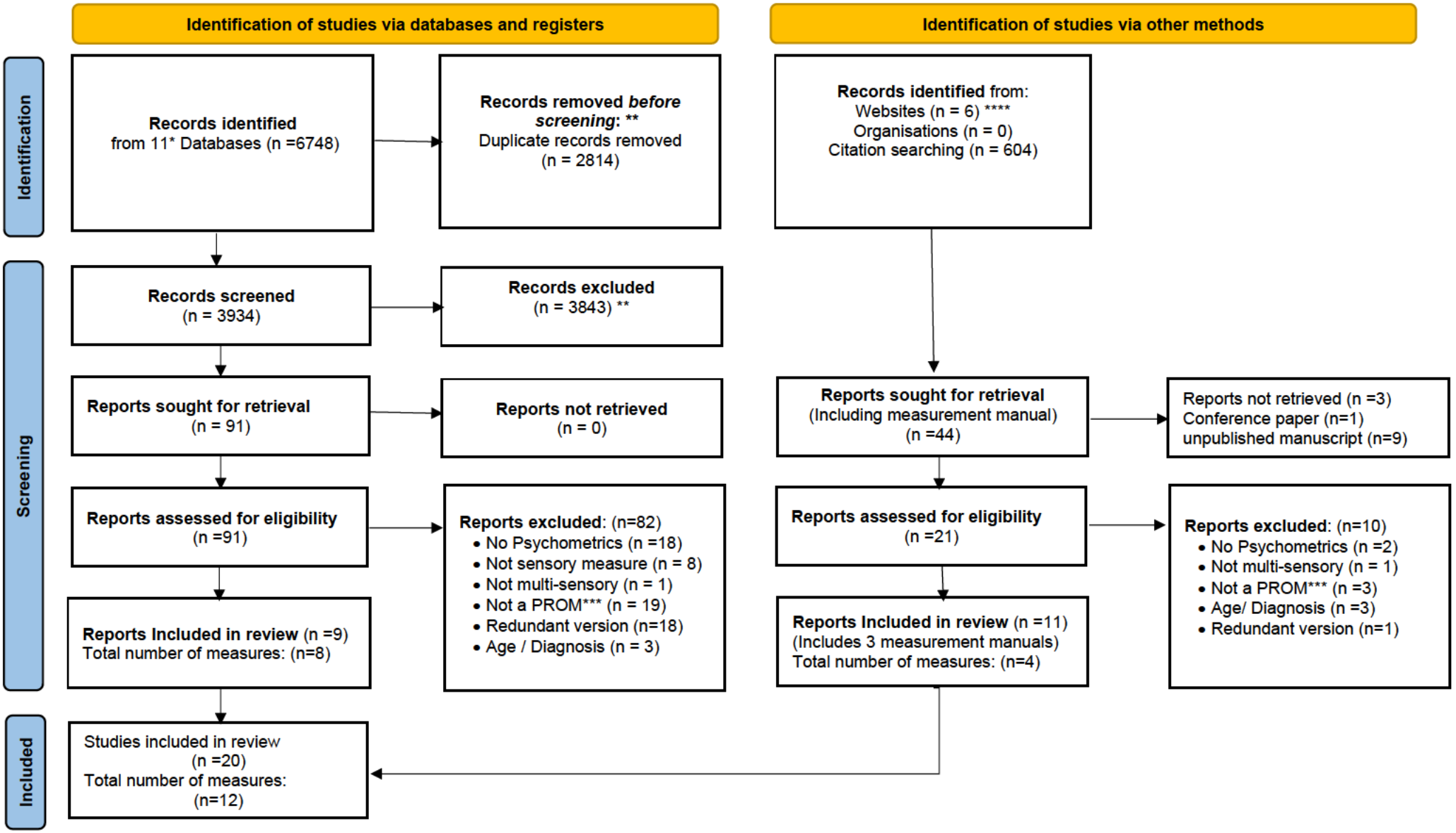


Figure S2: PRISMA 2020 Flowchart including all databases, registers, and other sources.

Table S1: PRISMA 2020 Checklist

Section and Topic	Item #	Checklist item	Location where item is reported
TITLE			
Title	1	Identify the report as a systematic review.	Page 1
ABSTRACT			
Abstract	2	See the PRISMA 2020 for Abstracts checklist.	Page 3
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of existing knowledge.	Page 5 & 6
Objectives	4	Provide an explicit statement of the objective(s) or question(s) the review addresses.	Page 6
METHODS			
Eligibility criteria	5	Specify the inclusion and exclusion criteria for the review and how studies were grouped for the syntheses.	Page 7
Information sources	6	Specify all databases, registers, websites, organisations, reference lists and other sources searched or consulted to identify studies. Specify the date when each source was last searched or consulted.	Pages 6, 7, 9 and 10
Search strategy	7	Present the full search strategies for all databases, registers and websites, including any filters and limits used.	Supplementary Table 2 and Supplementary Table 3
Selection process	8	Specify the methods used to decide whether a study met the inclusion criteria of the review, including how many reviewers screened each record and each report retrieved, whether they worked independently, and if applicable, details of automation tools used in the process.	Page 7 and 8
Data collection process	9	Specify the methods used to collect data from reports, including how many reviewers collected data from each report, whether they worked independently, any processes for obtaining or confirming data from study investigators, and if applicable, details of automation tools used in the process.	Page 7 and 8
Data items	10a	List and define all outcomes for which data were sought. Specify whether all results that were compatible with each outcome domain in each study were sought (e.g. for all measures, time points, analyses), and if not, the methods used to decide which results to collect.	Page 7 and 8 and Supplementary Tables 2 and 3
	10b	List and define all other variables for which data were sought (e.g. participant and intervention characteristics, funding sources). Describe any assumptions made about any missing or unclear information.	Page 6 and Page 14
Study risk of bias assessment	11	Specify the methods used to assess risk of bias in the included studies, including details of the tool(s) used, how many reviewers assessed each study and whether they worked independently, and if applicable, details of automation tools used in the process.	Pages 7,8 and 9
Effect measures	12	Specify for each outcome the effect measure(s) (e.g. risk ratio, mean difference) used in the synthesis or presentation of results.	Pages 7,8 and 9
Synthesis methods	13a	Describe the processes used to decide which studies were eligible for each synthesis (e.g. tabulating the study intervention characteristics and comparing against the planned groups for each synthesis (item #5)).	Figure 2
	13b	Describe any methods required to prepare the data for presentation or synthesis, such as handling of missing summary statistics, or data conversions.	Figure 1 and Page 7 and 23
	13c	Describe any methods used to tabulate or visually display results of individual studies and syntheses.	Figure 1, Table 1 and supplementary Table 4

Section and Topic	Item #	Checklist item	Location where item is reported
	13d	Describe any methods used to synthesize results and provide a rationale for the choice(s). If meta-analysis was performed, describe the model(s), method(s) to identify the presence and extent of statistical heterogeneity, and software package(s) used.	Figure 1, Pages 7, 8 and 9
	13e	Describe any methods used to explore possible causes of heterogeneity among study results (e.g. subgroup analysis, meta-regression).	Pages 7, 11, 16 and 17
	13f	Describe any sensitivity analyses conducted to assess robustness of the synthesized results.	Pages 7,8,and 9
Reporting bias assessment	14	Describe any methods used to assess risk of bias due to missing results in a synthesis (arising from reporting biases).	Pages 7, 8 and 9
Certainty assessment	15	Describe any methods used to assess certainty (or confidence) in the body of evidence for an outcome.	Pages 7, 8 and 9
RESULTS			
Study selection	16a	Describe the results of the search and selection process, from the number of records identified in the search to the number of studies included in the review, ideally using a flow diagram.	Page 9 and 10
	16b	Cite studies that might appear to meet the inclusion criteria, but which were excluded, and explain why they were excluded.	Page 10 and 11
Study characteristics	17	Cite each included study and present its characteristics.	Table 1
Risk of bias in studies	18	Present assessments of risk of bias for each included study.	Table 3
Results of individual studies	19	For all outcomes, present, for each study: (a) summary statistics for each group (where appropriate) and (b) an effect estimate and its precision (e.g. confidence/credible interval), ideally using structured tables or plots.	Table 1
Results of syntheses	20a	For each synthesis, briefly summarise the characteristics and risk of bias among contributing studies.	Table 3 and 5
	20b	Present results of all statistical syntheses conducted. If meta-analysis was done, present for each the summary estimate and its precision (e.g. confidence/credible interval) and measures of statistical heterogeneity. If comparing groups, describe the direction of the effect.	Table 4 and 6, Supplementary Table 7
	20c	Present results of all investigations of possible causes of heterogeneity among study results.	Table 3-6 and Supplementary Tables4,5 and 7
	20d	Present results of all sensitivity analyses conducted to assess the robustness of the synthesized results.	Table 3-6 and Supplementary Tables4,5 and 7
Reporting biases	21	Present assessments of risk of bias due to missing results (arising from reporting biases) for each synthesis assessed.	Table 3-6 and Supplementary Tables4,5 and 7
Certainty of	22	Present assessments of certainty (or confidence) in the body of evidence for each outcome assessed.	Table 4 and 6

PRISMA 2020 Checklist

Section and Topic	Item #	Checklist item	Location where item is reported
evidence			
DISCUSSION			
Discussion	23a	Provide a general interpretation of the results in the context of other evidence.	Page 17-20
	23b	Discuss any limitations of the evidence included in the review.	Page 20-21
	23c	Discuss any limitations of the review processes used.	Pages 20-21
	23d	Discuss implications of the results for practice, policy, and future research.	Pages 20-21
OTHER INFORMATION			
Registration and protocol	24a	Provide registration information for the review, including register name and registration number, or state that the review was not registered.	Page 6
	24b	Indicate where the review protocol can be accessed, or state that a protocol was not prepared.	Page 6
	24c	Describe and explain any amendments to information provided at registration or in the protocol.	NA
Support	25	Describe sources of financial or non-financial support for the review, and the role of the funders or sponsors in the review.	Page 2
Competing interests	26	Declare any competing interests of review authors.	Page 2
Availability of data, code and other materials	27	Report which of the following are publicly available and where they can be found: template data collection forms; data extracted from included studies; data used for all analyses; analytic code; any other materials used in the review.	Page 1

Table S2: Example of search strategy for Medline via Ovid

	Search Terms
Population: Terms related to age	adolescen*.mp. or Adolescent/ OR child*.mp. or exp Child/ OR p*ediatric.mp. OR preschool*.mp.OR school age.mp. OR teen*.mp. 0R youth*.mp.
	AND
Terms related to diagnosis	neurodevelopment* disorder*.mp. or exp Neurodevelopmental Disorders/ OR development* disorder*.mp. or exp developmental disorders/ OR neuropsych* disorder*.mp. OR Anxiety/ or anxiety.mp. OR depressive disorder*.mp. or Depressive Disorder/ OR exp Autism Spectrum Disorder/ OR autis*.mp. OR attention deficit disorder.mp. OR communication disorder*.mp OR intellect* disab*.mp. OR learning disorder*.mp. OR global developmental delay*.mp. OR Obsessive-Compulsive Disorder/ or obsessive compulsive disorder*.mp. OR intellectual development* disorder*.mp. OR exp Language Disorders/ or language disorder*.mp. OR tic disorder*.mp. OR Tourette Syndrome/ or tourette*.mp
	AND
Measure: Terms relating to sensory dysregulation	atypical sensory reactivity.mp. OR (process* adj1 (sensory or sense*)).mp. OR sensorimotor.mp. OR (sensory adj1 (dysregulation or defensiveness or discrimination or modulation or overreactivity or over reactivity or perception or reactivity or seeking)).mp. OR ((sensory or sense*) adj1 (overrespons* or over-respons*)).mp. OR somatic hypersensitivity.mp. OR somatosensory.mp.
	AND
Terms relating to the type of measure	assess*.mp. OR evaluat*.mp. OR checklist*.mp. or Checklist/ OR instrument*.mp. OR inventor*.mp. OR measure*.mp. OR profile*.mp. OR screen*.mp. OR survey*.mp. OR "Surveys and Questionnaires"/ OR tool*.mp. OR test*.mp. OR questionnaire*.mp. OR validation Stud*.mp
	AND
Measurement properties	alpha.mp. OR ceiling effect.mp. OR coefficien*.mp. OR computer adaptive test*.mp. OR concordanc*.mp. OR cronbach*.mp. OR cross cultural equivalence.mp. OR dimensionalit*.mp. OR differential item function*.mp. OR dif.mp. OR Discriminant Analysis/ OR discrimina*.mp. ORfactor analys?s.mp. or Factor Analysis, Statistical/ OR factor structure*.mp. OR finding*.mp. OR floor effect.mp. OR generali*.mp. OR individual variab*.mp. OR ((inter* or intra) adj1 (rater or tester or examiner* or assay or variab* or individ* or participant* or observ* or technic*)).mp. OR interrater.mp. OR intrarater.mp. OR intertester.mp. OR intratester.mp. OR interexaminer*.mp. OR intraexaminer*.mp. OR interassay.mp. OR intraassay.mp. OR intervariab*.mp. OR intravariab*.mp. OR interindivid*.mp. OR intraindivid*.mp. OR interparticipant*.mp. OR intraparticipant*.mp. OR interobserv*.mp. OR intraobserv*.mp. OR intertechnic*.mp. OR intratechnic*.mp. OR interscale correlation*.mp. OR internal consistency.mp. OR irt.mp. OR (item adj1 (bank* or correlation or discriminant or selection or reduction or response model*)).mp. OR kappa*.mp. OR meaningful change*.mp. OR ((minimal* detectable or minimal* important or minmum real) adj1 (concentration or change* or differen*)).mp. OR multitrait scaling analysis.mp. OR outcome*.mp. OR observer variation*.mp. OR precision.mp. OR psychometric*.mp. or Psychometrics/ OR precise value*.mp. OR quotient*.mp. OR rasch.mp. OR rate variabilit*.mp. OR reliability.mp. OR re test*.mp. OR retest*.mp. OR repeatab*.mp. OR (repeat* adj1 measur*).mp. OR strateg*.mp.OR reproducib*.mp. or "Reproducibility of Results"/ OR result*.mp. OR sensitiv*.mp. OR stability.mp. ORstandard error of measurement.mp. OR specific*.mp. or "Sensitivity or Specificity"/ OR subscal*.mp. OR valid*.mp. OR variab* analysis.mp.

Table S3: Search Strategies for all 11 databases used in this systematic review

Clinical Question	Population	Type of Measure	Measurement properties	Study Design
Management	Children and adolescents with a neurodevelopmental disorder	Sensory assessment measures/ screening tool / evaluation measures / outcome measures/ survey / questionnaire / profile/ test / inventory which reports on sensory dysregulation	Information on the psychometric; validity; reliability; responsiveness; sensitivity and specificity; reproducibility of results of various assessment	Any study design

Search terms – terms across columns combined with "AND"
No limits were set for any data of the database searches.

Medline via Ovid

Search Population	Intervention or Exposure	Outcome
Age: 1. adolescen*.mp. or Adolescent/ 2. child*.mp. or exp Child/ 3. p*ediatric.mp. 4. preschool*.mp. 5. school age.mp. 6. teen*.mp. 7. youth*.mp. 8. **1 OR 2 OR 3 OR 4 OR 5 OR 6 OR 7** **Diagnosis:** 9. neurodevelopment* disorder*.mp. or exp Neurodevelopmental Disorders/ 10. development* disorder*.mp. or exp developmental disorders/ 11. neuropsych* disorder*.mp. 12. Anxiety/ or anxiety.mp. 13. depressive disorder*.mp. or Depressive Disorder/ 14. exp Autism Spectrum Disorder/ or autis*.mp. 15. attention deficit disorder.mp. 16. communication disorder*.mp 17. intellect* disab*.mp. 18. learning disorder*.mp.	26. atypical sensory reactivity.mp. 27. (process* adj1 (sensory or sense*)).mp. 28. sensorimotor.mp. 29. (sensory adj1 (dysregulation or defensiveness or discrimination or modulation or overreactivity or over reactivity or perception or reactivity or seeking)).mp. 30. ((sensory or sense*) adj1 (overrespons* or over-respons*)).mp. 31. somatic hypersensitivity.mp. 32. somatosensory.mp. 33. **26 OR 27 OR 28 OR 29 OR 30 OR 31 OR 32** 34. assess*.mp. 35. evaluat*.mp. 36. checklist*.mp. or Checklist/ 37. instrument*.mp. 38. inventor*.mp. 39. measure*.mp. 40. profile*.mp.	49. alpha.mp. 50. ceiling effect.mp. 51. coefficien*.mp. 52. computer adaptive test*.mp. 53. concordanc*.mp. 54. cronbach*.mp. 55. cross cultural equivalence.mp. 56. dimensionalit*.mp. 57. differential item function*.mp. 58. dif.mp. 59. Discriminant Analysis/ 60. discrimina*.mp. 61. factor analys?s.mp. or Factor Analysis, Statistical/ 62. factor structure*.mp. 63. finding*.mp. 64. floor effect.mp. 65. generali*.mp. 66. individual variab*.mp. 67. ((inter* or intra) adj1 (rater or tester or examiner* or assay or variab* or individ* or participant* or observ* or technic*)).mp. 68. interrater.mp. 69. intrarater.mp. 70. intertester.mp.

19. global developmental delay*.mp.
20. Obsessive-Compulsive Disorder/ or obsessive compulsive disorder*.mp.
21. intellectual development* disorder*.mp.
22. exp Language Disorders/ or language disorder*.mp.
23. tic disorder*.mp.
24. Tourette Syndrome/ or tourette*.mp.
25. **9 OR 10 OR 11 OR 12 OR 13 OR 14 OR 15 OR 16 OR 17 OR 18 OR 19 OR 20 OR 21 OR 22 OR 24 OR 24**

41. screen*.mp.
42. survey*.mp.
43. "Surveys and Questionnaires"/
44. tool*.mp.
45. test*.mp.
46. questionnaire*.mp.
47. validation Stud*.mp.
48. **34 OR 35 OR 36 OR 37 OR 38 OR 39 OR 40 OR 41 OR 42 OR 43 OR 44 OR 45 OR 46 OR 47**

71. intratester.mp.
72. interexaminer*.mp.
73. intraexaminer*.mp.
74. interassay.mp.
75. intraassay.mp.
76. intervariab*.mp.
77. intravariab*.mp.
78. interindivid*.mp.
79. intraindivid*.mp.
80. interparticipant*.mp.
81. intraparticipant*.mp.
82. interobserv*.mp.
83. intraobserv*.mp.
84. intertechnic*.mp.
85. intratechnic*.mp.
86. interscale correlation*.mp.
87. internal consistency.mp.
88. irt.mp.
89. (item adj1 (bank* or correlation or discriminant or selection or reduction or response model*)).mp.
90. kappa*.mp.
91. meaningful change*.mp.
92. ((minimal* detectable or minimal* important or minmum real) adj1 (concentration or change* or differen*)).mp.
93. multitrait scaling analysis.mp.
94. outcome*.mp.
95. observer variation*.mp.
96. precision.mp.
97. psychometric*.mp. or Psychometrics/
98. precise value*.mp.
99. quotient*.mp.
100. rasch.mp.
101. rate variabilit*.mp.
102. reliability.mp.
103. re test*.mp.
104. retest*.mp.
105. repeatab*.mp.
106. (repeat* adj1 measur*).mp.

107. strateg*.mp.
108. reproducib*.mp. or "Reproducibility of Results"/
109. result*.mp.
110. sensitiv*.mp.
111. stability.mp.
112. standard error of measurement.mp.
113. specific*.mp. or "Sensitivity or Specificity"/
114. subscal*.mp.
115. valid*.mp.
116. variab* analysis.mp.
117. **49 OR 50 OR 51 OR 52 OR 53 OR 54 OR 55 OR 56 OR 57 OR 58 OR 59 OR 60 OR 61 OR 62 OR 63 OR 64 OR 65 OR 66 OR 67 OR 68 OR 69 OR 70 OR 71 OR 72 OR 73 OR 74 OR 75 OR 76 OR 77 OR 78 OR 79 OR 80 OR 81 OR 82 OR 83 OR 84 OR 85 OR 86 OR 87 OR 88 OR 89 OR 90 OR 91 OR 92 OR 93 OR 94 OR 95 OR 96 OR 97 OR 98 OR 99 OR 100 OR 101 OR 102 OR 103 OR 104 OR 105 OR 106 OR 107 OR 108 OR 109 OR 110 OR 111 OR 112 OR 113 OR 114 OR 115 OR 116**

118. **8 AND 25 AND 33 AND 48 AND 117**

PreMedline		
Search Population	**Intervention or Exposure**	**Outcome**
#1: adolescent* OR child* OR p*ediatric* OR preschool* OR "school age*" OR youth **#2:**"neurodevelopment* disorder*" OR "neuropsych* disorder*" OR "depressive disorder*" OR autis* OR "communicat* disorder*" OR "language disorder*" OR "learning disorder*" OR "global developmental delay*" OR "intellectual development* disorder*" OR "attention deficit disorder" OR 'attention deficit hyperactivity disorder" OR "obsessive compulsive disorder*" OR "tic* disorder*" OR tourette*	**#3:** "atypical sensory reactivity" OR "sens* process*" OR sensor*motor* OR "somatic hypersensitivity" OR somatosensory OR sensor* adj1 (dysregulation OR defensiveness OR discrimination OR "over respons*" OR overrespons* OR modulation OR overreactiv* OR "over reactiv*" OR perception* OR reactiv* OR seek*) **#4:** assess* OR evaluat* OR checklist* OR instrument* OR inventor* OR measur* OR profile* OR screen* OR survey* OR tool* OR test* OR questionnaire* OR "validation stud*"	**#5:** "computer adaptive testing" OR concordance OR cronbach* OR "cross cultural equivalen*" OR dimensionalit* OR "differential item function*" OR 7dif OR "discriminany analysis" OR discriminative OR "factor analys*s" OR "7factor structure*" OR finding* OR "ceiling effect" OR "floor effect" OR generali* OR "individual variab*" OR intrarater* OR interrater* OR intertester* OR intratester* OR interexaminer* OR intraexaminer* OR interassay* OR intraassay* OR interindividual* OR intraindividual* OR (inter OR intra) adj1 (rater* OR tester* OR examiner* OR assay* OR individual* OR participant* OR observer* OR technician*) OR "interval variability" OR interparticipant* OR intraparticipant* OR intraobserver* OR interobserver* OR intratechnician* OR intertechnician* OR "interscale correlation*" OR "internal consistenc*" OR kappa* OR "meaningful change*" OR specif* OR item adj1 (bank* OR correlation OR discriminant OR selection OR reduction OR "response model") **#6:** ("minimal* detectable" OR "minimal* important" OR "minimal* real") adj1 (change OR difference OR concentration) OR "multitrait scaling analysis" OR outcome* OR "observer variation*" OR precis* OR psychometric* OR "precise value*" OR quotient* OR rasch OR "rate variab*" OR strateg* OR "re test" OR retest OR result* OR repeatab* OR measur* OR reproducib* OR sensitiv* OR stability OR subscale* OR valid* OR "variability analysis" Use the 'combine queries' box to combine these individual searches: **#7. #5 OR #6** **#8. #1 AND #2 AND #3 AND #4 AND #7**

AMED via Ovid		
Search Population	**Intervention or Exposure**	**Outcome**
Age: 1. adolescen*.mp. or Adolescent/ 2. child*.mp. or exp Child/ 3. p*ediatric.mp. 4. preschool*.mp. 5. school age.mp. 6. teen*.mp. 7. youth*.mp. 8. **1 OR 2 OR 3 OR 4 OR 5 OR 6 OR 7** **Diagnosis:** 9. neurodevelopment* disorder*.mp. 10. Developmental Disabilities/ 11. development* disorder*.mp. 12. neuropsych* disorder*.mp. 13. Anxiety/ or anxiety.mp. 14. depressive disorder*.mp. or Depressive Disorder/ 15. autism.mp. 16. attention deficit disorder.mp. 17. communication disorder*.mp 18. intellect* disab*.mp. 19. learning disorder*.mp. 20. global developmental delay*.mp. 21. Obsessive-Compulsive Disorder/ or obsessive compulsive disorder*.mp. 22. intellectual development* disorder*.mp. 23. exp Language Disorders/ or language disorder*.mp. 24. tic disorder*.mp. 25. Tourette Syndrome/ or tourette*.mp. 26. **9 OR 10 OR 11 OR 12 OR 13 OR 14 OR 15 OR 16 OR 17 OR 18 OR 19 OR 20 OR 21 OR 22 OR 24 OR 24**	27. atypical sensory reactivity.mp. 28. (process* adj1 (sensory or sense*)).mp. 29. sensorimotor.mp. 30. (sensory adj1 (dysregulation or defensiveness or discrimination or modulation or overreactivity or over reactivity or perception or reactivity or seeking)).mp. 31. ((sensory or sense*) adj1 (overrespons* or over-respons*)).mp. 32. somatic hypersensitivity.mp. 33. somatosensory.mp. 34. **27 OR 28 OR 29 OR 30 OR 31 OR 32 OR 33** 35. assess*.mp. 36. evaluat*.mp. 37. checklist*.mp. or Checklist/ 38. instrument*.mp. 39. inventor*.mp. 40. measure*.mp. 41. profile*.mp. 42. screen*.mp. 43. survey*.mp. 44. "Surveys and Questionnaires"/ 45. tool*.mp. 46. test*.mp. 47. questionnaire*.mp. 48. validation Stud*.mp. 49. **35 OR 36 OR 37 OR 38 OR 39 OR 40 OR 41 OR 42 OR 43 OR 44 OR 45 OR 46 OR 47 OR 48**	50.alpha.mp. 51. ceiling effect.mp. 52. coefficien*.mp. 53.computer adaptive test*.mp. 54. concordanc*.mp. 55. cronbach*.mp. 56. cross cultural equivalence.mp. 57. dimensionalit*.mp. 58. differential item function*.mp. 59. dif.mp. 60. Discriminant Analysis/ 61. discrimina*.mp. 62. factor analys?s.mp. or Factor Analysis, Statistical/ 63. factor structure*.mp. 64. finding*.mp. 65. floor effect.mp. 66. generali*.mp. 67. individual variab*.mp. 68. ((inter* or intra) adj1 (rater or tester or examiner* or assay or variab* or individ* or participant* or observ* or technic*)).mp. 69. interrater.mp. 70. intrarater.mp. 71. intertester.mp. 72. intratester.mp. 73. interexaminer*.mp. 74. intraexaminer*.mp. 75. interassay.mp. 76. intraassay.mp. 77. intervariab*.mp. 78. intravariab*.mp. 79. interindivid*.mp. 80. intraindivid*.mp. 81. interparticipant*.mp. 82. intraparticipant*.mp. 83. interobserv*.mp.

84. intraobserv*.mp.
85. intertechnic*.mp.
86. intratechnic*.mp.
87.interscale correlation*.mp.
88. internal consistency.mp.
89.irt.mp.
90. (item adj1 (bank* or correlation or discriminant or selection or reduction or response model*)).mp.
91. kappa*.mp.
92. meaningful change*.mp.
93. ((minimal* detectable or minimal* important or minimum real) adj1 (concentration or change* or differen*)).mp.
94. multitrait scaling analysis.mp.
95. outcome*.mp.
96. observer variation*.mp.
97. precision.mp.
98. psychometric*.mp. or Psychometrics/
99. precise value*.mp.
100. quotient*.mp.
101. rasch.mp.
102. rate variabilit*.mp.
103. reliability.mp.
104. re test*.mp.
105. retest*.mp.
106. repeatab*.mp.
107. (repeat* adj1 measur*).mp.
108. strateg*.mp.
109. reproducib*.mp. or "Reproducibility of Results"/
110. result*.mp.
111. sensitiv*.mp.
112. stability.mp.
113. standard error of measurement.mp.
114. specific*.mp. or "Sensitivity or Specificity"/
115. subscal*.mp.
116. valid*.mp.
117. variab* analysis.mp.
118. **50 OR 51 OR 52 OR 53 OR 54 OR 55 OR 56 OR 57 OR 58 OR 59 OR 60 OR 61 OR 62 OR 63 OR 64 OR 65 OR 66 OR 67 OR**

68 OR 69 OR 70 OR 71 OR 72 OR 73 OR 74 OR 75 OR 76 OR 77 OR 78 OR 79 OR 80 OR 81 OR 82 OR 83 OR 84 OR 85 OR 86 OR 87 OR 88 OR 89 OR 90 OR 91 OR 92 OR 93 OR 94 OR 95 OR 96 OR 97 OR 98 OR 99 OR 100 OR 101 OR 102 OR 103 OR 104 OR 105 OR 106 OR 107 OR 108 OR 109 OR 110 OR 111 OR 112 OR 113 OR 114 OR 115 OR 116 OR 117

119. **8 AND 26 AND 34 AND 49 AND 118**

PsycInfo via Ovid		
Search Population	**Intervention or Exposure**	**Outcome**
Age:	29. atypical sensory reactivity.mp.	52. alpha.mp.
1. adolescen*.mp. or Adolescent/	30. (process* adj1 (sensory or sense*)).mp.	53. ceiling effect.mp.
2. child*.mp.	31. sensorimotor.mp.	54. coefficien*.mp.
3. p?ediatric.mp.	32. (sensory adj1 (dysregulation or defensiveness or discrimination or modulation or overreactivity or over reactivity or perception or reactivity or seeking)).mp.	55.computer adaptive test*.mp.
4. preschool*.mp.		56. concordanc*.mp.
5. school age.mp.		57. cronbach*.mp.
6. teen*.mp.		58. cross cultural equivalence.mp.
7. youth*.mp.		59. dimensionalit*.mp.
8. 1 OR 2 OR 3 OR 4 OR 5 OR 6 OR 7		60. differential item function*.mp.
	33. ((sensory or sense*) adj1 (overrespons* or over-respons*)).mp.	61. dif.mp.
Diagnosis:		62. Discriminant Analysis/
9. Developmental Disabilities/	34. somatic hypersensitivity.mp.	63. discrimina*.mp.
10. development* disorder*.mp	35. somatosensory.mp.	64. factor analys?s.mp. or Factor Analysis, Statistical/
11. neurodevelopment* disorder*.mp. or exp Neurodevelopmental Disorders/	**36. 29 OR 30 OR 31 OR 32 OR 33 OR 34 OR 35**	65. factor structure*.mp.
12. neuropsych* disorder*.mp.		66. finding*.mp.
13. Anxiety/ or anxiety.mp.	37. assess*.mp.	67. floor effect.mp.
14. exp Autism Spectrum Disorder/ or autis*.mp.	38. evaluat*.mp.	68. generali*.mp.
15. attention deficit disorder.mp.	39. checklist*.mp. or Checklist/	69. individual variab*.mp.
16. Communication disorders/ or communication disorder*.mp	40. instrument*.mp.	70. ((inter* or intra) adj1 (rater or tester or examiner* or assay or variab* or individ* or participant* or observ* or technic*)).mp.
17. depressive disorder*.mp. or Depressive Disorder/	41. inventor*.mp.	
	42. measure*.mp.	71. interrater.mp.
18. Intellectual developmental disorders/ or intellectual development* disorder*.mp.	43. profile*.mp.	72. intrarater.mp.
	44. screen*.mp.	73. intertester.mp.
19. intellect* disab*.mp.	45. survey*.mp.	74. intratester.mp.
20. Delayed development/ or delayed development.mp.	46. "Surveys and Questionnaires"/	75. interexaminer*.mp.
	47. tool*.mp.	76. intraexaminer*.mp.
21. Learning Disorders/ or learning disorder*.mp.	48. test*.mp.	77. interassay.mp.
22. global developmental delay*.mp.	49. questionnaire*.mp.	78. intraassay.mp.
23. Obsessive-Compulsive Disorder/ or obsessive compulsive disorder*.mp	**50.** validation Stud*.mp.	79. intervariab*.mp.
	51. 37 OR 38 OR 39 OR 40 OR 41 OR 42 OR 43 OR 44 OR 45 OR 46 OR 47 OR 48 OR 49 OR 50	80. intravariab*.mp.
24. exp specific Language Disorders/ or language disorder*.mp.		81. interindivid*.mp.
		82. intraindivid*.mp.
25. tic disorder*.mp.		83. interparticipant*.mp.
26. Tourette Syndrome/ or tourette*.mp.		84. intraparticipant*.mp.
		85. interobserv*.mp.

27. Nervous system disorders/ or nervous* system disorder*.mp.

28. 9 OR 10 OR 11 OR 12 OR 13 OR 14 OR 15 OR 16 OR 17 OR 18 OR 19 OR 20 OR 21 OR 22 OR 24 OR 24 OR 25 OR 26 OR 27

86. intraobserv*.mp.
87. intertechnic*.mp.
88. intratechnic*.mp.
89. interscale correlation*.mp.
90. internal consistency.mp.
91. irt.mp.
92. (item adj1 (bank* or correlation or discriminant or selection or reduction or response model*)).mp.
93. kappa*.mp.
94. meaningful change*.mp.
95. ((minimal* detectable or minimal* important or minmum real) adj1 (concentration or change* or differen*)).mp.
96. multitrait scaling analysis.mp.
97. outcome*.mp.
98. observer variation*.mp.
99. precision.mp.
100. psychometric*.mp. or Psychometrics/
101. precise value*.mp.
102. quotient*.mp.
103. rasch.mp.
104. rate variabilit*.mp.
105. reliability.mp.
106. re test*.mp.
107. retest*.mp.
108. repeatab*.mp.
109. (repeat* adj1 measur*).mp.
110. strateg*.mp.
111. reproducib*.mp. or "Reproducibility of Results"/
112. result*.mp.
113. sensitiv*.mp.
114. stability.mp.
115. standard error of measurement.mp.
116. specific*.mp. or "Sensitivity or Specificity"/
117. subscal*.mp.
118. valid*.mp.
119. variab* analysis.mp.
120. **52 OR 53 OR 54 OR 55 OR 56 OR 57 OR 58 OR 59 OR 60 OR 61 OR 62 OR 63 OR 64 OR 65 OR 66 OR 67 OR 68 OR 69 OR**

70 OR 71 OR 72 OR 73 OR 74 OR 75 OR 76 OR 77 OR 78 OR 79 OR 80 OR 81 OR 82 OR 83 OR 84 OR 85 OR 86 OR 87 OR 88 OR 89 OR 90 OR 91 OR 92 OR 93 OR 94 OR 95 OR 96 OR 97 OR 98 OR 99 OR 100 OR 101 OR 102 OR 103 OR 104 OR 105 OR 106 OR 107 OR 108 OR 109 OR 110 OR 111 OR 112 OR 113 OR 114 OR 115 OR 116 OR 117 OR 118 OR 119

121. 8 AND 28 AND 36 AND 51 AND 120

Embase via Ovid		
Search Population	**Intervention or Exposure**	**Outcome**
Age:	29.atypical sensory reactivity.mp.	52. alpha.mp.
1. adolescen*.mp. or Adolescent/	30.(process* adj1 (sensory or sense*)).mp.	53. ceiling effect.mp.
2. child*.mp. or exp Child/	31.sensorimotor.mp.	54. coefficien*.mp.
3. p?ediatric.mp.	32.(sensory adj1 (dysregulation or defensiveness or discrimination or modulation or overreactivity or over reactivity or perception or reactivity or seeking)).mp.	55.computer adaptive test*.mp.
4. preschool*.mp.		56. concordanc*.mp.
5. school age.mp.		57. cronbach*.mp.
6. teen*.mp.		58. cross cultural equivalence.mp.
7. youth*.mp.		59. dimensionalit*.mp.
8. **1 OR 2 OR 3 OR 4 OR 5 OR 6 OR 7**	33.((sensory or sense*) adj1 (overrespons* or over-respons*)).mp.	60. differential item function*.mp.
Diagnosis:	34.somatic hypersensitivity.mp.	61. dif.mp.
9. Developmental Disabilities/	35.somatosensory.mp.	62. Discriminant Analysis/
10. development* disorder*.mp	36.**29 OR 30 OR 31 OR 32 OR 33 OR 34 OR 35**	63. discrimina*.mp.
11. neurodevelopment* disorder*.mp. or exp Neurodevelopmental Disorders/		64. factor analys?s.mp. or Factor Analysis, Statistical/
12. neuropsych* disorder*.mp.		65. factor structure*.mp.
13. Anxiety/ or anxiety.mp.	37.assess*.mp.	66. finding*.mp.
14. exp Autism Spectrum Disorder/ or autis*.mp.	38.evaluat*.mp.	67. floor effect.mp.
15. attention deficit disorder.mp.	39.checklist*.mp. or Checklist/	68. generali*.mp.
16. Communication disorders/ or communication disorder*.mp	40.instrument*.mp.	69. individual variab*.mp.
17. depressive disorder*.mp. or Depressive Disorder/	41.inventor*.mp.	70. ((inter* or intra) adj1 (rater or tester or examiner* or assay or variab* or individ* or participant* or observ* or technic*)).mp.
18. Intellectual developmental disorders/ or intellectual development* disorder*.mp.	42.measure*.mp.	71. interrater.mp.
19. intellect* disab*.mp.	43.profile*.mp.	72. intrarater.mp.
20. Delayed development/ or delayed development.mp.	44.screen*.mp.	73. intertester.mp.
21. Learning Disorders/ or learning disorder*.mp.	45.survey*.mp.	74. intratester.mp.
22. global developmental delay*.mp.	46."Surveys and Questionnaires"/	75. interexaminer*.mp.
23. Obsessive-Compulsive Disorder/ or obsessive compulsive disorder*.mp	47.tool*.mp.	76. intraexaminer*.mp.
24. exp Language Disorders/ or language disorder*.mp.	48.test*.mp.	77. interassay.mp.
25. tic disorder*.mp.	49.questionnaire*.mp.	78. intraassay.mp.
	50.validation Stud*.mp.	79. intervariab*.mp.
	51.**37 OR 38 OR 39 OR 40 OR 41 OR 42 OR 43 OR 44 OR 45 OR 46 OR 47 OR 48 OR 49 OR 50**	80. intravariab*.mp.
		81. interindivid*.mp.
		82. intraindivid*.mp.
		83. interparticipant*.mp.
		84. intraparticipant*.mp.
		85. interobserv*.mp.

26. Tourette Syndrome/ or tourette*.mp.
27. Nervous system disorders/ or nervous* system disorder*.mp.
28. **9 OR 10 OR 11 OR 12 OR 13 OR 14 OR 15 OR 16 OR 17 OR 18 OR 19 OR 20 OR 21 OR 22 OR 24 OR 24 OR 25 OR 26 OR 27**

86. intraobserv*.mp.
87. intertechnic*.mp.
88. intratechnic*.mp.
89. interscale correlation*.mp.
90. internal consistency.mp.
91. irt.mp.
92. (item adj1 (bank* or correlation or discriminant or selection or reduction or response model*)).mp.
93. kappa*.mp.
94. meaningful change*.mp.
95. ((minimal* detectable or minimal* important or minimum real) adj1 (concentration or change* or differen*)).mp.
96. multitrait scaling analysis.mp.
97. outcome*.mp.
98. observer variation*.mp.
99. precision.mp.
100. psychometric*.mp. or Psychometrics/
101. precise value*.mp.
102. quotient*.mp.
103. rasch.mp.
104. rate variabilit*.mp.
105. reliability.mp.
106. re test*.mp.
107. retest*.mp.
108. repeatab*.mp.
109. (repeat* adj1 measur*).mp.
110. strateg*.mp.
111. reproducib*.mp. or "Reproducibility of Results"/
112. result*.mp.
113. sensitiv*.mp.
114. stability.mp.
115. standard error of measurement.mp.
116. specific*.mp. or "Sensitivity or Specificity"/
117. subscal*.mp.
118. valid*.mp.
119. variab* analysis.mp.
120. **52 OR 53 OR 54 OR 55 OR 56 OR 57 OR 58 OR 59 OR 60 OR 61 OR 62 OR 63 OR 64 OR 65 OR 66 OR 67 OR 68 OR 69 OR**

70 OR 71 OR 72 OR 73 OR 74 OR 75 OR 76 OR 77 OR 78 OR 79 OR 80 OR 81 OR 82 OR 83 OR 84 OR 85 OR 86 OR 87 OR 88 OR 89 OR 90 OR 91 OR 92 OR 93 OR 94 OR 95 OR 96 OR 97 OR 98 OR 99 OR 100 OR 101 OR 102 OR 103 OR 104 OR 105 OR 106 OR 107 OR 108 OR 109 OR 110 OR 111 OR 112 OR 113 OR 114 OR 115 OR 116 OR 117 OR 118 OR 119

121. **8 AND 28 AND 36 AND 51 AND 120**

CINAHL		
Search Population	**Intervention or Exposure**	**Outcome**
Age:	S31. "atypical sensory reactivity"	S64. (MH "Coefficient Alpha") OR "Alpha"
S1. MH adolescent*	S32. "sensory process*"	S65. (MH "Factor Analysis") OR "factor Analysis"
S2. (MH "Child+") OR "child*"	S33. (MH "Sensory Motor Integration") OR "sensorimotor"	S66. (MH "Data Analysis, Statistical+") OR "statistical"
S3. "P#ediatric"	S34. "sensory dysregulation"	S67. "ceiling effect"
S4. (MH "Child, Preschool") OR "preschool*"	S35. (MH "Sensory Defensiveness") OR "sensory defensiveness"	S68. (MH "Pearson's Correlation Coefficient") OR (MH "Coefficient Alpha") OR (MH "Correlation Coefficient+") OR (MH "Spearman's Rank Correlation Coefficient") OR (MH "Kuder-Richardson Coefficient") OR (MH "Intraclass Correlation Coefficient") OR (MH "Kappa Statistic") OR (MH "Step-Wise Multiple Regression") OR (MH "Multiple Logistic Regression") OR "coefficient*"
S5. "school age"	S36. "sensory discrimination"	S69. "coefficient of variation"
S6. (MH "Adolescence+") OR "teen*"	S37. "sensory overrespons*"	S70. (MH "Computerized Adaptive Testing") OR "computer adaptive testing"
S7. "youth*"	S38. (MH "Hypersensitivity, Immediate+") OR (MH "Hypersensitivity, Delayed") OR (MH "Hypersensitivity+") OR "somatic hypersensitivity"	S71. "Concordance*"
S8. S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7	S39. "sensory modulation"	S72. "cronbach*"
	S40. "sensory over* reactivity"	S73. (MH "Ethnological Research") OR "cross-cultural equivalence"
Diagnosis:	S41."sensory perception"	S74. "dimensionality*"
S9. ‘‘neurodevelopmental disorder*’’	S42."sensory reactivity"	S75. (MH "Differential Item Functioning") OR "differential item functioning"
S10. ‘‘neurodevelopment* disorder*’’	S43."sensory seeking"	S76. "DIF"
S11. ‘‘developmental disorder*’’	S44.(MH "Somatosensory Disorders+") OR (MH "Proprioception+") OR "somatosensory"	S77. (MH "Discriminant Analysis") OR "discriminant analysis"
S12. MH Child development* disorder*	S45. "sensory sensitivity"	S78. "discriminative"
S13. neuropsychiatric disorder*	**S46. S31 OR S32 OR S33 OR S34 OR S35 OR S36 OR S37 OR S38 OR S39 ORS 40 OR S41 OR S42 OR S43 OR S44 OR S45**	S79. "factor*analys?s"
S14. (MH "Anxiety+") OR "anxiety" OR (MH "Anticipatory Anxiety") OR (MH "Social Anxiety Disorders") OR (MH "Anxiety Disorders+") OR (MH "Separation Anxiety") OR (MH "Generalized Anxiety Disorder")		S80. "factor structure*"
S15.(MH "Affective Disorders, Psychotic+") OR "depressive disorder"	S47. (MH "Outcome Assessment") OR "assess*"	S81. "findings"
S16. (MH "Attention Deficit Hyperactivity Disorder") OR "attention deficit disorder"	S48. (MH "Evaluation+") OR "evaluat*"	S82. "floor effect"
S17. (MH "Autistic Disorder") OR "autism Spectrum Disorder"	S49. ((MH "Occupational Therapy Assessment") OR "occupational therapy assessment" OR (MH "Pediatric Occupational Therapy")	S83. "generali?a*"
S18. (MH "Communicative Disorders+") OR "Communication Disorders"	S50. "Checklist"	S84. "Individual variability"
S19.(MH "Intellectual Disability+") OR "Intellectual Disability" OR (MH "Developmental Disabilities")	S51. (MH "Instrument Validation") OR (MH "Instrument Scaling+") OR "instrument*"	S85. (MH "Interrater Reliability") OR "int?r*rater*"
S20. "intellectual* disab*"	S52. "instrumentation" OR (MH "Instrument Validation")	S86. "int?r*tester*"
S21. (MH "Learning Disorders+") OR "Learning Disorders")	S53. "inventor*" OR (MH "Inventories")	S87. "intra*examiner*"
S22."global developmental delay*"		S88. "inter*assay*"
S23.(MH "Obsessive-Compulsive Disorder+") OR		S89. "interval variability"

(MH "Compulsive Personality Disorder") OR "obsessive compulsive disorder"

S24."intellectual development* disorder*"

S25. (MH "Language Disorders+") OR "Language Disorders"

S26. (MH "Pervasive Developmental Disorder-Not Otherwise Specified") OR (MH "Child Development Disorders, Pervasive+") OR "pervasive developmental disorder not otherwise specified"

S27. (MH "Child Behavior Disorders+") OR "child behavior disorders"

S28. "tic disorder*"

S29. (MH "Tourette Syndrome") OR "Tourette*"

S30. S9 OR S10 OR S11 OR S12 OR S13 OR S14 OR S15 OR S16 OR S17 OR S18 OR S19 OR S20 OR S21 OR S22 OR S24 OR S24 OR S25 OR S26 OR S27 OR S28 OR S29

S54. "measure*"

S55. "profile*"

S56. "screen*"

S57. (MH "Surveys+") OR "survey*"

S58. "tool*"

S59. "test*"

S60. "questionnaire*"

S61. (MH "Surveys+") OR "survey*"

S62. (MH "Validation Studies") OR "validation Stud*"

S63. 47 OR 48 OR 49 OR 50 OR 51 OR 52 OR 53 OR 54 OR 55 OR 56 OR 57 OR 58 OR 59 OR 60 OR 61 OR 62

S90. "intra*assay*"

S91. "intra-assay*"

S92. "intra*individual*"

S93. "intra*participant*"

S94. "inter*observer*"

S95. "intra*observer"

S96. "inter*examiner*"

S97. "interscale correlation*"

S98. "inter*individual*"

S99. "inter*participant*"

S100.(MH "Internal Consistency+") OR "internal consistency"

S101. "IRT"

S102. "item bank"

S103. "item correlation" OR (MH "Item-Total Correlations")

S104. "item discriminant"

S105. "item selection"

S106. "item reduction"

S107. "item response model"

S108. (MH "Kappa Statistic") OR "kappa*"

S109. "meaningful change*"

S110. "minimal detectable concentration"

S111. "minimal* important change"

S112. "minimal* important difference"

S113. (MH "Outcome Assessment") OR "outcome*"

S114. "observer variation*"

S115. (MH "Precision") OR "precision"

S116. (MH "Psychometrics") OR "psychometric*"

S117. "precise values"

S118. "quotient*"

S119. (MH "Rasch Analysis")

S120. "rate variability"

S121. (MH "Reliability+") OR "reliability" OR (MH "Reliability and Validity+") OR (MH "Test-Retest Reliability") OR (MH "Intrarater Reliability") OR (MH "Equipment Reliability")

S122. "strategy"

S123. (MH "Test-Retest Reliability") OR "re#test*" OR (MH "Rescreening")
S124. "result*"
S125. (MH "Repeated Measures") OR "repeatab*"
S126. "repeated measure*"
S127. "strateg*"
S128. (MH "Reproducibility of Results") OR "reproducib*"
S129. (MH "Sensitivity and Specificity") OR "sensitivity"
S130. (MH "Stability+") OR "stability"
S131. (MH "Measurement Error+") OR (MH "Descriptive Statistics") OR "standard error of measurement"
S132. "specificity"
S133. "subscale*"
S134. (MH "Predictive Validity") OR (MH "Reliability and Validity+") OR (MH "Internal Validity") OR (MH "Instrument Validation") OR (MH "Face Validity") OR (MH "External Validity") OR (MH "Discriminant Validity") OR (MH "Criterion-Related Validity+") OR (MH "Consensual Validity") OR (MH "Concurrent Validity") OR (MH "Validation Studies") OR (MH "Qualitative Validity+") OR (MH "Construct Validity+") OR "valid*"
S135. "variability analysis"
S136. (MH "Item Analysis") OR "item analysis"
S137. (MH "Test Construction") OR "test construct"
S138. S64 OR S65 OR S66 OR S67 OR S68 OR S69 OR S70 OR S71 OR S72 OR S73 OR S74 OR S75 OR S76 OR S77 OR S78 OR S79 OR S80 OR S81 OR S82 OR S83 OR S84 OR S85 OR S86 OR S87 OR S88 OR S89 OR S90 OR S91 OR S92 OR S93 OR S94 OR S95 OR S96 OR S97 OR S98 OR S99 OR S100 OR S101 OR S102 OR S103 OR S104 OR S105 OR S106 OR S107 OR S108 OR S109 OR S110 OR S111 OR S112 OR S113 OR S114 OR S115 OR S116 OR S117 OR S118 OR S119 OR S120 OR S121 OR S122 OR S123 OR S124 OR

S125 OR S126 OR S127 OR S128 OR S129 OR S130 OR S131 OR S132 OR S133 OR S134 OR S135 OR S136 OR S137

S139. S8 AND S30 AND S46 AND S63 AND S138

Scopus		
Search Population	**Intervention or Exposure**	**Outcome**
#1: adolescent* OR child* OR p?ediatric* OR preschool* OR "school age*" OR youth **#2:**"neurodevelopment* disorder*" OR "neuropsych* disorder*" OR "depressive disorder*" OR autis* OR "communicat* disorder*" OR "language disorder*" OR "learning disorder*" OR "global developmental delay*" OR "intellectual development* disorder*" OR attention W/3 "deficit disorder" OR "obsessive compulsive disorder*" OR "tic* disorder*" OR tourette*	**#3:** "atypical sensory reactivity" OR "sens* process*" OR sensor?motor* OR "somatic hypersensitivity" OR somatosensory OR sensor* W/1 (dysregulation OR defensiveness OR discrimination OR "over respons*" OR overrespons* OR modulation OR overreactiv* OR "over reactiv*" OR perception* OR reactiv* OR seek*) **#4:** assess* OR evaluat* OR checklist* OR instrument* OR inventor* OR measur* OR profile* OR screen* OR survey* OR tool* OR test* OR questionnaire* OR "validation stud*"	**#5:** "computer adaptive testing" OR concordanc* OR cronbach* OR "cross cultural equivalen*" OR dimensionalit* OR "differential item function*" OR 7dif OR "discriminany analysis" OR discriminative OR "factor analys?s" OR "7factor structure*" OR finding* OR "ceiling effect" OR "floor effect" OR generali* OR "individual variab*" OR intrarater* OR interrater* OR intertester* OR intratester* OR interexaminer* OR intraexaminer* OR interassay* OR intraassay* OR interindividual* OR intraindividual* OR (inter OR intra) W/1 (rater* OR tester* OR examiner* OR assay* OR individual* OR participant* OR observer* OR technician*) OR "interval variability" OR interparticipant* OR intraparticipant* OR intraobserver* OR interobserver* OR intratechnician* OR intertechnician* OR "interscale correlation*" OR "internal consistenc*" OR kappa* OR "meaningful change*" OR specif* OR item W/1 (bank* OR correlation OR discriminant OR selection OR reduction OR "response model") **#6:** ("minimal* detectable" OR "minimal* important" OR "minimal* real") W/1 (change OR difference OR concentration) OR "multitrait scaling analysis" OR outcome* OR "observer variation*" OR precis* OR psychometric* OR "precise value*" OR quotient* OR rasch OR "rate variab*" OR strateg* OR "re test" OR retest OR result* OR repeatab* OR measur* OR reproducib* OR sensitiv* OR stability OR subscale* OR valid* OR "variability analysis"

Use the ‘combine queries’ box to combine these individual searches:

#7. #5 OR #6
#8. #1 AND #2 AND #3 AND #4 AND #7

Web of science		
Search Population	**Intervention or Exposure**	**Outcome**
#1: adolescent* OR child* OR p$ediatric* OR preschool* OR "school age*" OR youth **#2:** "neurodevelopment* disorder*" OR "neuropsych* disorder*" OR "depressive disorder*" OR autis* OR "communicat* disorder*" OR "language disorder*" OR "learning disorder*" OR "global developmental delay*" OR "intellectual development* disorder*" OR (attention NEAR/3 "deficit disorder") OR "obsessive compulsive disorder*" OR "tic* disorder*" OR tourette*	**#3:** "atypical sensory reactivity" OR "sens* process*" OR sensor$motor* OR "somatic hypersensitivity" OR somatosensory OR sensor* NEAR/1 (dysregulation OR defensiveness OR discrimination OR "over respons*" OR overrespons* OR modulation OR overreactiv* OR "over reactiv*" OR perception* OR reactiv* OR seek*) **#4:** assess* OR evaluat* OR checklist* OR instrument* OR inventor* OR measur* OR profile* OR screen* OR survey* OR tool* OR test* OR questionnaire* OR "validation stud*"	**#5:** "computer adaptive testing" OR concordanc* OR cronbach* OR "cross cultural equivalen*" OR dimensionalit* OR "differential item function*" OR 7dif OR "discriminany analysis" OR discriminative OR "factor analys$s" OR "7factor structure*" OR finding* OR "ceiling effect" OR "floor effect" OR generali* OR "individual variab*" OR intrarater* OR interrater* OR intertester* OR intratester* OR interexaminer* OR intraexaminer* OR interassay* OR intraassay* OR interindividual* OR intraindividual* OR ((inter OR intra) NEAR/1 (rater* OR tester* OR examiner* OR assay* OR individual* OR participant* OR observer* OR technician*)) OR "interval variability" OR interparticipant* OR intraparticipant* OR intraobserver* OR interobserver* OR intratechnician* OR intertechnician* OR "interscale correlation*" OR "internal consistenc*" OR kappa* OR "meaningful change*" OR specif* OR (item NEAR/1 (bank* OR correlation OR discriminant OR selection OR reduction OR "response model")) **#6:** (("minimal* detectable" OR "minimal* important" OR "minimal* real") NEAR/1 (change OR difference OR concentration)) OR "multitrait scaling analysis" OR outcome* OR "observer variation*" OR precis* OR psychometric* OR "precise value*" OR quotient* OR rasch OR "rate variab*" OR strateg* OR "re test" OR retest OR result* OR repeatab* OR measur* OR reproducib* OR sensitiv* OR stability OR subscale* OR valid* OR "variability analysis"

Use the ‘combine queries’ box to combine these individual searches:

#7. #5 OR #6
#8. #1 AND #2 AND #3 AND #4 AND #7

InformIt		
Search Population	**Intervention or Exposure**	**Outcome**
#1: adolescent* OR child* OR p?ediatric* OR preschool* OR "school age*" OR youth AND **#2:** "neurodevelopment* disorder*" OR "neuropsych* disorder*" OR "depressive disorder*" OR autis* OR "communicat* disorder*" OR "language disorder*" OR "learning disorder*" OR "global developmental delay*" OR "intellectual development* disorder*" OR (attention %3 "deficit disorder") OR "obsessive compulsive disorder*" OR "tic* disorder*" OR tourette*	**AND** **#3:** "atypical sensory reactivity" OR "sens* process*" OR sensor*motor* OR "somatic hypersensitivity" OR somatosensory OR sensor* %1 dysregulation OR sensor* %1 defensiveness OR sensor* %1 discrimination OR sensor* %1 "over respons*" OR sensor* %1 overrespons* OR sensor* %1 modulation OR sensor* %1 overreactiv* OR sensor* %1 "over reactiv*" OR sensor* %1 perception OR sensor* %1 reactiv* OR sensor* %1 seek* AND **#4:**assess* OR evaluat* OR checklist* OR instrument* OR inventor* OR measur* O R profile* OR screen* OR survey* OR too l* OR test* OR questionnaire* OR "valida tion stud*"	**AND** **#5:**("computer adaptive testing" OR concordanc* OR cronbach* OR "cross cultural equivalen*" OR dimensionalit* OR "differential item function*" OR 7dif OR "discriminany analysis" OR discriminative OR "factor analys*2s" OR "7factor structure*" OR finding* OR "ceiling effect" OR "floor effect" OR generali* OR "individual variab*" OR intrarater* OR interrater* OR intertester* OR int ratester* OR interexaminer* OR intraexaminer* OR interassay * OR intraassay* OR interindividual* OR intraindividual* ORinter %1 rater* OR inter %1 tester* OR inter %1 examiner* OR inter %1 assay OR inter %1 individual OR inter %1 participant OR inter %1 observer* OR inter %1 technician* OR intra %1 rater* OR intra %1 tester* OR intra %1 examiner* OR intra %1 assay* OR intra %1 individual* OR intra %1 participant* OR intra %1 observer* OR intra %1 technician* OR "interval variability" OR interparticipant* OR intraparticipant* OR intra observer* OR interobserver* OR intratechnician* OR intertec hnician* OR "interscale correlation*" OR "internal consistenc*" OR kappa* OR "meaningful change*" OR specif* OR item %1 bank* OR item %1 correlation OR item %1 discrimination OR item %1 selection OR item %1 reduction OR item %1 "response model") AND ("minimal* detectable" %1 change OR "minimal* detectable" %1 difference OR "minimal* detectable" %1 concentration OR "minimal* important" %1 change OR "minimal* important" %1 difference OR "minimal* important" %1 concentration OR "minimal* real" %1 change OR "minimal* real" %1 difference OR concentration OR "multitrait scaling analysis" OR outcome* OR "observer variation*" OR

precis* OR psychometric* OR "precise value*" OR quotient* OR rasch OR "rate variab*" OR strateg* OR "re test" OR retest OR result* OR repeatab* OR measur* OR reproducib* OR sensitiv* OR stability OR subscale* OR valid* OR "variability analysis")

Use the 'combine queries' box to combine these individual searches:

#7. #5 OR #6
#8. #1 AND #2 AND #3 AND #4 AND #7

Complementary and Alternative Medicine via Pubmed		
Search Population	**Intervention or Exposure**	**Outcome**
#1: Adolescent[MeSH] OR Child[MeSH] OR adolescent* OR child* OR p?ediatric* OR preschool* OR "school age*" OR youth **#2:** Neurodevelopmental Disorders [MeSH] OR developmental disorders[MeSH] OR Anxiety[MeSH] OR Depressive Disorder [MeSH] OR Autism Spectrum Disorder [MeSH] OR Obsessive-Compulsive Disorder [MeSH] OR Language Disorders [MeSH] OR Tourette Syndrome [MeSH] OR"neurodevelopment* disorder*" OR neuropsych* disorder*" OR "depressive disorder*" OR autis* OR "communicat* disorder*" OR "language disorder*" OR "learning disorder*" OR "global developmental delay*" OR "intellectual development* disorder*" OR (attentionW/3 "deficit disorder") OR "obsessive compulsive disorder*" OR "tic* disorder*" OR tourette*	**#3:** "atypical sensory reactivity" OR "sens* process*" OR sensor*motor* OR "somatic hypersensitivity" OR somatosensory OR (sensor*W/3(dysregulation OR defensiveness OR discrimination OR "over respons*" OR overrespons* OR modulation OR overreactiv* OR "over reactiv*" OR perception* OR reactiv* OR seek*)) **#4:** checklist*.mp. or Checklist[MeSH] OR "Surveys and Questionnaires"[MeSH] OR assess* OR evaluat* OR checklist* OR instrument* OR inventor* OR measur* OR profile* OR screen* OR survey* OR tool* OR test* OR questionnaire* OR "validation stud*"	**#5:** Discriminant Analysis[MeSH] OR Factor Analysis, Statistical [MeSH] OR Psychometrics[MeSH] OR "Reproducibility of Results"[MeSH] OR "Sensitivity or Specificity"[MeSH] OR "computer adaptive testing" OR concordanc* OR cronbach* OR "cross cultural equivalen*" OR dimensionalit* OR "differential item function*" OR 7dif OR "discriminany analysis" OR discriminative OR "factor analys*2s" OR "7factor structure*" OR finding* OR "ceiling effect" OR "floor effect" OR generali* OR "individual variab*" OR intrarater* OR interrater* OR intertester* OR intratester* OR interexaminer* OR intraexaminer* OR interassay* OR intraassay* OR interindividual* OR intraindividual* OR ((inter OR intra)W/3"(rater* OR tester* OR examiner* OR assay* OR individual* OR participant* OR observer* OR technician*)") OR "interval variability" OR interparticipant* OR intraparticipant* OR intraobserver* OR interobserver* OR intratechnician* OR intertechnician* OR "interscale correlation*" OR "internal consistenc*" OR kappa* OR "meaningful change*" OR specif* OR (itemW/3 (bank* OR correlation OR discriminant OR selection OR reduction OR "response model")) **#6:** ("minimal* detectable" OR "minimal* important" OR "minimal* real") W/3(change OR difference OR concentration) OR "multitrait scaling analysis" OR outcome* OR "observer variation*" OR precis* OR psychometric* OR "precise value*" OR quotient* OR rasch OR "rate variab*" OR strateg* OR "re test" OR retest OR result*

OR
repeatab* OR measur* OR reproducib* OR sensitiv* OR stability OR subscale* OR valid* OR "variability analysis"

Use the 'combine queries' box to combine these individual searches:

#7. #5 OR #6
#8. #1 AND #2 AND #3 AND #4 AND #7

Cochrane Library		
Search Population	**Intervention or Exposure**	**Outcome**
Age: 1. MeSH descriptor: [Adolescent] 2. adolescen*: 3. MeSH descriptor: [Child] 4. child* 5. p*ediatric 6. preschool* 7. school age 8. teen* 9. youth* **10.{OR #1-#9}** Diagnosis: 11.MeSH descriptor: [Neurodevelopmental Disorders] 12.neurodevelopment* disorder* 13.development* disorder* 14.MeSH descriptor: [Learning Disabilities] 15.neuropsych* disorder* 16.MeSH descriptor: [Anxiety] 17. anxiety 18.depressive disorder* 19.MeSH descriptor: [Depressive Disorder] 20.MeSH descriptor: [Autism Spectrum Disorder] 21. autis* 22.attention deficit disorder* 23.communication disorder* 24.intellect* disab* 25.learning disorder* 26.global developmental delay* 27.MeSH descriptor: [Obsessive-Compulsive Disorder] 28.obsessive compulsive disorder* 29.intellectual development* disorder*	36.atypical sensory reactivity 37.(process* NEAR/1 (sensory or sense*)) 38.sensorimotor* 39.(sensory NEAR/1 (dysregulation or defensiveness or discrimination or modulation or overreactivity or over reactivity or perception or reactivity or seeking)) 40.((sensory or sense*) NEAR/1 (overrespons* or over-respons*))* 41.somatic hypersensitivity* 42.somatosensory* **43.{OR #36-#42}** 44.assess* 45.evaluat* 46.checklist* 47. MeSH descriptor: [Checklist] 48.instrument* 49.inventor* 50.measure* 51.profile* 52.screen* 53.survey* 54.MeSH descriptor: [Surveys and Questionnaires] 55.tool* 56.test* 57.questionnaire* 58.validation Stud* **59.{OR #44-#58}**	60. alpha 61. ceiling effect* 62. coefficien* 63. computer adaptive test* 64. concordanc* 65. cronbach* 66. cross cultural equivalence* 67. dimensionalit* 68. differential item function* 69. dif* 70. MeSH descriptor: [Discriminant Analysis] 71. discrimina* 72. factor analys?s 73. MeSH descriptor: [Factor Analysis, Statistical] 74. factor structure* 75. finding* 76. floor effect* 77. generali* 78. individual variab* 79. ((inter* or intra) NEAR/1 (rater or tester or examiner* or assay or variab* or individ* or participant* or observ* or technic*)) 80. interrater 81. intrarater 82. intertester 83. intratester 84. interexaminer 85. intraexaminer* 86. interassay 87. intraassay 88. intervariab* 89. intravariab* 90. interindivid* 91. intraindivid*

30.MeSH descriptor: [Language Disorders]
31.language disorder*
32.tic disorder*
33.MeSH descriptor: [Tourette Syndrome]
34.Tourette*
35.{OR #11-#34}

92. interparticipant*
93. intraparticipant*
94. interobserv*
95. intraobserv*
96. intertechnic*
97. intratechnic*
98. interscale correlation*
99. internal consistency*
100. irt*
101. (item NEAR/ (bank* or correlation or discriminant or selection or reduction or response model*))*
102. kappa*
103. meaningful change*
104. ((minimal* detectable or minimal* important or minmum real) NEAR/1 (concentration or change* or differen*))
105. multitrait scaling analysis
106. outcome*
107. observer variation*
108. precision
109. psychometric*
110. MeSH descriptor: [Psychometrics]
111. precise value*
112. quotient*
113. rasch
114. rate variabilit*
115. reliability
116. re test*
117. retest*
118. repeatab*
119. (repeat* adj1 measur*)*
120. strateg*
121. reproducib*
122. MeSH descriptor: [Reproducibility of Results]
123. result*
124. sensitiv*
125. stability
126. standard error of measurement

127. specific*
128. subscal*
129. valid*
130. variab* analysis
131. {OR #60-#130}

132. (#10 AND #35 AND #43 AND #59 AND #131)

Table S4: COSMIN criteria for good measurement properties

Measurement property	Rating[1]	Criteria
Structural validity	+	**Classic Test Theory (CTT):** Confirmatory factor analysis (CFA): Comparative fit index (CFI) or Tucker-Lewis index (TLI) or comparable measure > 0.95 or RMSEA < 0.06 or SRMR <0.8[2] **Item response theory (IRT) /Rasch** No violation of unidimensionality[3]: CFI or TLI or comparable measure > 0.95 or Root Mean Square Error of Approximation (RMSEA) <0.06 or Standardized root mean residuals (SRMR) <0.08 AND No violation of local independence: residual correlations among the items after controlling for the dominant factor < 0.20. or Q3's <0.37 AND No violation of monotonicity: adequate looking graph or item scalability > 0.30 AND Adequate model fit: IRT: χ^2 > 0.01 Rasch: infit and outfit mean squares ≥ 0.5 and ≤ 1.5 or Z-standardized values > - 2 and < 2
	?	CTT: Not all information for '+' reported IRT /Rasch: Model fir not reported
	-	Criteria for '+' not met
Internal consistency	+	At least low evidence[4] for sufficient structural validity[5] AND Cronbach's alpha(s) ≥ 0.70 for each unidimensional scale or subscale[6]
	?	Criteria for "At least low evidence[4] for sufficient structural validity[5]" not met
	-	At least low evidence[4] for sufficient structural validity[5] AND Cronbach's alpha(s) < 0.70 for each unidimensional scale or subscale[6]
Reliability	+	Interclass correlation coefficient (ICC) or weighted Kappa ≥ 0.70
	?	ICC or weighted Kappa not reported
	-	ICC or weighted Kappa < 0.70
Measurement error	+	Smallest detectable change (SDC) or limits of agreement (LoA) < minimal important change (MIC[5])
	?	MIC not defined
	-	SDC or LoA > MIC[5]
Hypotheses testing for construct validity	+	The result is in accordance with the hypothesis[7]
	?	No hypothesis was defined (by the review team)
	-	The result is not in accordance with the hypothesis[7]
Cross-cultural validity/ measurement invariance	+	No important differences were found between-group factors (such as age, gender, language) in multiple group factor analysis or no important DIF for group factors (McFadden's R^2 < 0.02)
	?	No multiple group factor analysis or DIF analysis performed
	-	Important differences between group factors or DIF was found
Criterion validity	+	Correlation with reported gold standard ≥ 0.70 or AUC ≥ 0.70
	?	Not all information for '+'
	-	Correlation with reported gold standard < 0.70 or AUC < 0.70

Table S5: Definitions of the four different ratings for the quality of evidence

Quality of evidence	Definition	Grade Lower if:
High	We are very confident that the true measurement property lies close to that of the estimate* of the measurement property.	Risk of bias: -1 Serious -2 Very serious -3 Extremely serious
Moderate	We are moderately confident in the measurement property estimate: the true measurement property is likely to be close to the estimate of the measurement property, but there is a possibility that it is substantially different.	Inconsistency: -1 Serious -2 very serious
Low	Our confidence in the measurement property estimate is limited: the true measurement property may be substantially different from the estimate of the measurement property.	Imprecision: -1 Total n=50-100 -2 Total n<50
Very Low	We have very little confidence in the measurement property estimate: the true measurement property is likely to be substantially different from the estimate of the measurement property	Indirectness -1 Serious -2 Very serious

Table S6: Description of all included studies evaluated in this systematic review.

Measure	Acronym	# of studies	Reference	Purpose of study	Study population	Age (range [R] and/or Mean [M] Standard deviation [SD] and Gender details
Assessment of Sensory Processing and Executive Functions in Childhood	EPYFEI	1	Romero-Ayuso (2018) (47)	The aim of this study was to determine the psychometric properties of the "Assessment of Sensory Processing and Executive Functions in Childhood".	**n=383** Spanish children with a neurodevelopmental disorder (including ADHD (n=95, 5.5%); ASD (n=84, 4.8%); LSD (n=106, 6.1%); DD (n=15, 0.9%) & other neurodevelopmental disorders (n=83, 4.8%). n=1,349 Spanish neuro-typically developing children.	**Age**: Total sample: R=3-11yrs; M=6.6; SD=3.39 **Gender:** Total sample male: n=884 (51%)
Child Sensory Profile-2	CSP2	1	Dunn (2014) (41)*	Assessment manual	**n= 297** Clinical sample (DD=11, ASD=78, ADHD=96, Dual Dx =24, Learning delay=45, Gifted= 18, ID=9, Downs =9, English second language=7). n= 697 TD children	**Age**: Total sample: R= 3-14:11 yrs.; M=NR; SD=NR **Gender**: Clinical sample for only the CSP2 not provide. TD sample: male n=348 (49.9%) and female n=349 (50.1%)
Knickerbocker Sensorimotor History Questionnaire	KSHQ	1	Carrasco (1990) (48)	This study was part of a larger project to investigate the reliability of the Knickerbocker Sensorimotor History Questionnaire.	**n=20** Parents completing measure were predominately Caucasian with 2-8 yrs. of a college education.	**Age:** Total Parent sample: R=25-40 yrs.; M=NR; SD=NR Total sample Children: R=2-8 yrs.; M=NR; SD=NR **Gender:** Not stated
Participation and Sensory Environment Questionnaire–Home Scale	PSEQ-H	3	Pfeiffer (2018) (54)	The goal of this study was to develop a pool of items for an instrument that assesses parent perspectives concerning the impact of the sensory environment on participation in daily activities for their young children with ASD.	**n=35** Parents and caregivers interviewed had children between the ages of 2 to 7 yrs. with a diagnosis of ASD.	**Age:** Total Parents sample: R= 20-59 yrs.; M=NR; SD=NR Total Children sample: R=2-7 yrs.; M=4.5 yrs.; SD=1.45 **Gender** Parents: mothers n=33; male guardian n=1 Children: female n=6
			Pfeiffer (2018) (55)	A cross-sectional study was completed to determine internal consistency, test-retest reliability, and examine item distribution.	**n=167** Children with ASD and >70 scores on the GARS-3 (Gilliam Autism Rating) Scale-3. n= 137 TD children. *	**Age**: Total sample: R=2-7 yrs.; M=4.94 yrs.; SD=NR **Gender:** Children with ASD: male n=127 (76%); female n=39 (23%) TD children: male n=72 (53%); female n=65 (47%)

Table S6: Description of all included studies evaluated in this systematic review.

			Bevan (2020) (56)	To describe the psychometric evaluation of the PSEQ–H, including the tool's structural validity; item difficulty, discrimination, and bias; reliability; and construct validity.	**n=167** Children with ASD and >70 scores on the GARS-3 (Gilliam Autism Rating) Scale-3. n= 137 TD children*.	**Age:** Total sample: R=2-7 yrs.; M=4.94 yrs.; SD=NR **Gender:** Children with ASD: male n=127 (76%); female n=39 (23%) TD children: male n=72 (53%); female n=65 (47%)
Participation and Sensory Environment Questionnaire–Community Scale	PSEQ-C	1	Pfeiffer (2019) (57)	The purpose of this study was to examine the reliability, namely the internal consistency and test-retest reliability of the P–SEQ: Community Scales.	**n=141** Children with ASD and >70 scores on the GARS-3 (Gilliam Autism Rating) Scale-3. n= 161 Children who scored ≤70 on the GARS-3 were put in the neurotypical subgroup.	**Age:** Total sample: R=2-7 yrs.; M=NR; SD=NR **Gender:** Children with ASD male: n=113 (81%); female n=27 (19%) TD children: male n=84 (52%); female n=77 (48%)

Measure	Acronym	# of studies	Reference	Purpose of study	Study population	Age (range [R] and/or Mean [M] Standard deviation [SD] and Gender details
Sensory Behavior Questionnaire	SBQ	1	Neil (2017) (58)	This study examined the reliability and validity of the Sensory Behavior Questionnaire, a parent-report scale designed to assess frequency and impact of sensory behaviors in autistic children.	**n=66** Children with ASD. **n=70** TD children.	**Age:** Children with ASD: R=6.82-16.46 yrs.; M=10.28; SD=2.50 Children TD: R=6.-17.59 yrs.; M=9.90; SD=2.61 **Gender:** 32) Children with ASD: Male n=57, female n=9 TD children: Male n=36; Female n=34
Sensory Experiences Questionnaire version 3	SEQ-3.0	1	Ausderau (2013) (49)	The main purpose of this study is to provide empirical validation for the Sensory Experiences Questionnaire Version 3.0 with children with ASD	**n=1,307** Children with ASD ((ASD= (63 %), Asperger's= (22.1%), PDD-NOS= (24,4%))	**Age:** Total sample: R=2-12 yrs.; M=NR; SD=NR **Gender:** Male = 82.3% of sample
Sensory Processing Measure Home	SPM Home	6	Parham (2007) (42)	Assessment manual	**n=345** (Clinical sample) Children receiving occupational therapy (OT) intervention. **n=1051** (Standardized sample) Children at full-day kindergarten programs & sixth graders attending elementary schools (not middle schools). Children with mild academic or behavioral problems were not excluded to reflect the base rate of the mild problems in the general population.	**Age:** Clinical sample: R=5-13 yrs.; M=NR; SD=NR Standardized sample: R= 5-12 yrs.; M=NR; SD=NR **Gender:** Clinical sample: Male: n=253 (73.3%), female n= 92 (26.7%) Standardized sample: Male: n= 547 (52%), female n=504 (48%)

Table S6: Description of all included studies evaluated in this systematic review.

		Dugas (2018) (72)	This article documents the convergent validity of the Sensory Profile (SP) and the Sensory Processing Measure (SPM)–Home Form for children with ASD.	**n=34** Children with diagnostic criteria associated with ASD, Asperger's syndrome, or PDD-NOS.	**Age:** Total sample: R=5-8 yrs.; M=72.91 months; SD=4.50 **Gender**: Male= n=28 (82.4), female =n=6 (17.6%)
		Brown (2010) (73)	To investigate the convergent validity between the Sensory Profile, the Sensory Profile School Companion, and the Home and Main Classroom Forms of the Sensory Processing Measure.	**n=30** Mothers of a group of TD children aged 5-10 years.	**Age:** Total Parents sample: R= 18-56+ yrs.; M=NR; SD=NR Total Children sample: R=5-10 yrs.; M=NR; SD=NR **Gender:** Male n=14 (46.7 5), female n=16 (53.3%)
		Brown (2010) (71)	The aim of the study was to investigate the reliability (internal consistency and inter-rater reliability) of the Sensory Profile, the Sensory Profile School Companion (SPSC), and the Home and Main Classroom Forms of the Sensory Processing Measure (SPM) in a cross-cultural setting.	**n=60** Mothers (n=30) and fathers (n=30) of a group of children ages 5 to 10 years.	**Age:** Total Parents sample: R= 18-56+ yrs.; M=NR; SD=NR Total sample Children: R=5-10 yrs.; M=NR; SD=NR **Gender:** Total Parents sample: Male n=30, female n=30. Total sample Children: Male: 14 (46%), female n=16 (53,3%)

Measure	Acronym	# of studies	Reference	Purpose of study	Study population	Age (range [R] and/or Mean [M] Standard deviation [SD] and Gender details
Sensory Processing Measure Home (Continued)	SPM Home		Lai (2011) (69)	This study aimed to examine the psychometric properties of the Sensory Processing Measure-Hong Kong Chinese version (SPM-HKC).	**n=100** Chinese children with ASD **n=542** TD Chinese children	**Age:** Children with ASD: R=5-12 yrs.; M=87 months; SD= 22months TD children: R=5-12 yrs.; M=93 months; SD=29 months. **Gender**: Children with ASD: Male: 78%, female: 12% TD sample: Male: 51.3%, female: 48.7%
Sensory Processing Measure Home	**SPM Home**		Ahmad (2020) (60)	To enhance the applicability and meaningfulness of SPM Home Form for the Malay speaking population, a study that focuses on translating, adapting, and validating the SPM Home Form into the Malay language was conducted.	**n=30** Parents with children with ASD between 5 to 12-year-old, Malaysian citizens.	**Age**: Total Parents sample: R= 30-49 yrs.; M=NR; SD=NR Total Children sample: R=5-12 yrs.; M=NR; SD=NR **Gender:** Total Parents sample: Male n=8 (26.7%), female n=22 (73.3%)

Table S6: Description of all included studies evaluated in this systematic review.

Sensory Processing Measure Preschool Home	SPM-P Home	2	Ecker & Parham (2010) (51)	Assessment manual	**n=242** (Clinical sample) Preschool children receiving occupational therapy (OT) intervention. **n=651** (Standardized sample) Two- to five-year-old children. Five-year-olds were included if they hadn't started kindergarten yet. Children with mild academic or behavioural difficulties were not excluded to reflect the mild problems in the general population.	**Age**: Clinical sample: R=2-5yrs; M=NR; SD=NR Standardized sample: R= 2-5 yrs.; M=NR; SD=NR **Gender:** Clinical sample: Male n=167 (69%); female n=75 (31.0) Standardized sample: Male: n=319 (49%), female n=332 (51.0%)
			Alkhalifah (2019) (70)	This study assessed the psychometric properties of the Sensory Processing Measure–Preschool Home Form (SPM-P-Home) when used in English with a population of English-speaking Saudi participants.	**n=16** Caregivers of children with ASD. **n=40** Caregiver of TD children	**Age:** Children with ASD: R=2-5yrs; M=4.26 yrs.; SD=1.28 TD sample: R=2-5yrs; M=3.20 yrs.; SD=1.07 **Gender:** Children with ASD: Male n=13, female n=3 TD Sample: Male n=18, female n=22
Sensory Processing Scale Inventory	SP Scale Inventory	1	Schoen (2017) (52)	The purpose of this paper is to report on the development and current psychometric properties of the SP Scale Inventory.	**n=267** Clinical sample (Children with sensory challenges without ASD, CP, Down syndrome, genetic abnormalities, or other DD). **n=140** TD.	**Age:** Clinical sample: R= 4-18 yrs.; M=6.96; SD=2.18 TD sample: R= 4-18 yrs.; M=8.13; SD=2.39 **Gender:** Clinical sample: Male: 69%, Female= 31% TD sample: Male: 53%, female: 47%
Sensory Processing and Self-Regulation Checklist	SPSRC	1	Lai (2019) (50)	This study is aimed at evaluating the psychometric properties of the SPSRC and examine the patterns of self-regulation and sensory processing in children with and without ASD.	**n=78** Chinese children with ASD. **n=997** TD Chinese children	**Age:** Children with ASD: R=43-106 months; M=NR; SD=NR TD sample: R=37-107 months; M=NR; SD=NR **Gender:** Children with ASD: Male: 79.5% and female: 20.5% TD sample: Male: 52.5% and female: 46.9%

Measure	Acronym	# of studies	Reference	Purpose of study	Study population	Age (range [R] and/or Mean [M] Standard deviation [SD] and Gender details
Short Sensory Profile-2	SSP-2	2	Dunn (2014) (41)*	Assessment manual	Same as SP2	Same as SP2
Short Sensory Profile-2	SSP-2-PL		Chojnicka (2019) (53)	This paper presents the psychometric characteristics of the Polish version of the SSP-2 verified on a relatively large	**n=310** Children with ASD (Autism (n=201); PDD-NOS / Asperger syndrome (n=109)).	**Age**: Total sample: R= 3-14 yrs.; M=NR; SD=NR **Gender**: Total sample: Male: 57%, female: 43% Children with ASD: Male 80%, female: 20% Non ASD group: Male: 43%, female: 57%

Table S6: Description of all included studies evaluated in this systematic review.

	sample of Polish children aged 3 to 14 years.	**n= 264** neurodevelopmental disorders. **n=656** TD children.	Non-spectrum	TD Group: Male: 57%, female: 43%

Table S7: Details of the characteristics of the 12 included measures.

Measure	Acronym	# of studies	Reference	Year Developed	Target population	Purpose of Measure	Number of items	Duration	Response format	Domains Assessed	Original language	Available translation
Assessment of Sensory Processing and Executive Functions in Childhood	EPYFEI	1	Romero-Ayuso (2018)(47)	2018	3 -11 years	Initial screening tool to assist with planning.	34	15 min	Five-point Likert scale	Five Domains: i) Attention, working memory, and initiation of actions (11 items); ii) global sensory processing, particularly tactile, proprioceptive, and vestibular information (7 items); iii) emotional and behavioural self-regulation (5 items); iv) organization, execution, supervision, and problem-solving in activities of daily living (6 items); and v) inhibitory control (5 items).	Spanish	-
Child Sensory Profile-2	CSP2	1	Dunn (2014)(41)	2014	3-14:11 years	Provides a standard method for professionals to document children's sensory processing patterns in the context of a child's everyday life.	86	15-20 min	Five-point Likert scale	Sensory pattern scores: i) Seeking/ seeker; ii) avoiding/ avoider; iii) sensitivity/ sensor; iv) registration/ bystander based on Dunn's Sensory Processing Framework. Sensory system scores: i) auditory; ii) visual; iii) touch; iv) movement; v) body position; vi) oral. Behavioral scores: i) Behavioral; ii) conduct, iii) social emotional; iv) attentional.	English	Spanish version (Dunn 2014) (41)
Knickerbocker Sensorimotor History Questionnaire	KSHQ	1	Carrasco (1990)(48)	1990	Not stated	Used to gather a child's sensory and motor history through a parent informant.	Not stated	Not stated	Not stated	Seven Domains include i) olfactory; ii) auditory; iii) visual; iv) motor organisation; v) tactile; vi) social adjustment, and vii) academically related questions	English/ Hebrew unclear	-
Participation and Sensory Environment Questionnaire– Home Scale	PSEQ-H Scale	3	Pfeiffer (2018)(54) Pfeiffer (2018)(55) Bevan (2020)(56)	2018	2-7 years	To assess the impact of the sensory environment on the participation of children within the home activities.	15	Not stated	Five-point Likert scale	Three sub-domains: i) Impact of sensory environment on participation in daily activities; ii) Amount of parent/caregiver effort required to support participation, and iii) Perceived meaningfulness / importance of the daily activity.	English	-

Table S7: Details of the characteristics of the 12 included measures.

Measure	Acronym	# of studies	Reference	Year Developed	Target population	Purpose of Measure	Number of items	Duration	Response options	Domains Assessed	Original language	Available translation
Participation and Sensory Environment Questionnaire– Community Scale	PSEQ-C Scale	2*	Pfeiffer (2018)(54) Pfeiffer (2019)(57)	2018	2-7 years	Designed to examine the impact of the sensory environment on participation within community activities.	19	Not stated	Five-point Likert scale	Three sub-domains: i) Impact of sensory environment on participation in community activities; ii) Amount of parent/caregiver effort required to support participation, and iii) Perceived meaningfulness/ importance of community activity.	English	-
Sensory Behavior Questionnaire	SBQ	1	Neil (2017)(58)	2009	6-17 years Cognitively able children with ASD	Designed to assess frequency and impact of sensory behaviors in autistic children.	50	Not stated	Six-point Likert scale	Domains: i) auditory; ii) visual; iii) movement (vestibular and proprioceptive); iv) tactile; v) oral motor (including gustatory olfactory) processing, and vi) general reactions and organisation.	English	-
Sensory Experiences Questionnaire version 3	SEQ-3.0	1	Ausderau (2013)(49)	2013	2-12 years with ASD	Designed to measure behavioral responses to naturally occurring sensory stimuli in the context of everyday situations in children with ASD and/or DD in social and non-social contexts.	97	15-20 min	Five-point Likert scale	Five modality categories: i) auditory; ii) visual; iii) tactile; iv) gustatory/olfactory and v) vestibular/ proprioceptive) across two contexts (social and non-social).	English	-

Table S7: Details of the characteristics of the 12 included measures.

Sensory Processing Measure Home	SPM Home form	6	Parham (2007)(51) Dugas (2018)(72) Brown (2010)(73) Brown (2010)(71)	2007	5-12 years	The SPM assesses social participation, praxis, and sensory processing issues of children.	75	15-20 min	Four-point Likert scale	Domains: i) social participation; ii) vision; iii) hearing; iv) touch; v) body awareness; vi) balance and motion; vii) planning and ideas; viii) total sensory systems; and five additional items on taste and smell processing.	English	SPM-HKC (Chinese) Lai (2011)(69) SPM-MV (Malay version) Ahmad (2020)(60)

Measure	Acronym	# of studies	Reference	Year Developed	Target population	Purpose of Measure	Number of items	Duration	Response options	Domains Assessed	Original language	Available translation
Sensory Processing Measure Preschool-Home	SPM-P-Home	2	Ecker & Parham (2010)(51)	2010	2-5 years	Assesses the degree the child is dysfunctional in each sensory system, including praxis and social involvement facilitates and distinguishes between sensory and behavioral difficulties.	75	15-20 min	Four-point Likert scale	Eight areas: i) social participation; ii) vision; iii) hearing; iv) Touch; v) Taste and Smell; vi) Body Awareness; vii) Balance and Motion; viii) Planning and Ideas (praxis).	English	Saudi Population using English version (Alkhalifah (2019)(70)
Sensory Processing Scale Inventory	SP Scale Inventory	1	Schoen (2017)(52)	2010	4-18 years	To characterise patterns of sensory modulation in children with an intellectual or developmental disability.	76	15-20 min	Binary scoring system used	Eight sensory domains: i) tactile; ii) auditory; iii) visual; iv) smell; v) taste; vi) vestibular; vii) proprioception and viii) interoception	English	-

Table S7: Details of the characteristics of the 12 included measures.

Sensory Processing and Self-Regulation Checklist	SPSRC	1	Lai (2019)(50)	2013	3-8 years with/without ASD	An instrument for the examination of sensory processing and self-regulation difficulties in children.	93	Not stated	Five-point Likert scale	Part 1: Tests self-regulation: i) physiological; ii) social/cognitive /emotional and iii) facing changes /challenges. Part 2: Tests sensory processing: i) auditory ii) visual; iii) tactile; iv) gustatory/olfactory; v) vestibular and vi) proprioceptive.	Chinese	-
Short Sensory Profile-2	SSP-2	2	Dunn (2014)(41)	2014	3-14:11 years	Provide quick information for screening and research programs.	34	5-10 min	Five-point Likert scale	Two Domains: i) Sensory Processing and ii) Behavioral Responses. In addition, scores are calculated for the four quadrants according to the theoretical model by Dunn.	English	SSP-2 -PL (Polish) Chojnicka (2019) (53)

Table S8: List of all excluded sensory measures from this systematic review and rationale for exclusion

Measure	Reference	Reason for exclusion
Assessment measure has been superseded		
Evaluation of Sensory Processing (ESP)	Johnson-Ecker (2000) (61)	The ESP was superseded by Sensory Processing Measure
Sensory Experiences Questionnaire (SEQ)	Baranek (2006) (62); Little (2011)(63)	The SEQ was superseded by Sensory Experiences Questionnaire version 3
Sensory Over-Responsivity Scale (SenSOR)	Schoen (2008) (64)	The SenSor was superseded by the Sensory Processing Scale Inventory
Sensory Profile (SP)	Dunn (1999) (41)	The SP was superseded by Sensory Profile 2nd edition
Short Sensory Profile (SSP)	McIntosh, D. N., Miller. J., & Shyu, V. (1999); Williams et al. (2018) (65)	The SSP was superseded by Short Sensory Profile 2nd edition
Assessment measures and psychometric publications not being published		
Expanded sensory profile	-	The expanded sensory profile and papers related to any psychometrics of the assessment tool have not been published.
Sensory History Questionnaire for Pre-schooler	-	The Sensory History Questionnaire for Pres-schoolers is referenced in a book chapter by DeGngi (2000) but no further information about the assessment measure or psychometrics of the tool appears to be published.
Sensory Processing Assessment for Young	-	Sensory Processing Assessment for Young was referenced by Baranek, G.T. (1999B) in an unpublished manuscript submitted to the University of North Carolina, Chapel Hill, NC
Sensory Problem Questionnaire	-	The Sensory Problem Questionnaire was referenced by Edelson, S.M. (1992) through the Center for the study of Autism but the assessment measure nor any psychometric studies appear to be published.
Sensory Questionnaire	-	Sensory Questionnaire was referenced by Boyd, BA. and Baranek, G.T. in an unpublished manuscript submitted to the University of North Carolina, Chapel Hill, NC
Sensory Supplement Questionnaire	-	The Sensory Supplement Questionnaire was presented by Parsons, H.N., Tignor, J., Beers, S., & Baranek, G.T. (2001) at The American Occupational Therapy Association's 81st Annual Conference and Expo, Philadelphia, PA. The conference abstract was retrieved by not further information on the assessment measure or psychometrics of the measure could be retrieved.

Table S8: List of all excluded sensory measures from this systematic review and rationale for exclusion

Measure	Reference	Reason for exclusion
Assessment measure not for age range of systematic review		
Adolescent/ Adult SP (AASP)	Brown, C, & Dunn, W. (2002) (70)	No subgroup analysis is available for age groups in the systematic review (3-18 yrs).
Brain Body Center Sensory Scale (BBCSS)	Kolacz (2018) (66)	No subgroup analysis is available for age groups in the systematic review (3-18 yrs).
Sensory Processing Quotient	Tavassoli, T., Hoekstra, R. A., & Baron-Cohen, S. (2014) (67)	The Sensory Processing Quotient is an assessment measure used with adults and not paediatrics, therefore this measure has been excluded from this systematic review.

Table S9: Quality of measure development results using COSMIN Risk of Bias checklist

Measure	Study (Ref)	Structural validity	Internal consistency	Cross-cultural validity	Reliability	Measurement error	Criterion validity	Hypothesis Testing for construct validity	
								Convergent validity	Discriminant validity
Assessment of sensory processing and executive function in childhood (EPYFEI)	Romero-Ayuso (2018) (47)	**?**	**+**	**NR**	**+**	**NR**	**-**	**-**	**-**
Child Sensory Profile 2 (CSP2)	Dunn (2014) (41)	**NR**	**+**	**-**	**+**	**+**	**?**	**+**	**+**
Knickerbocker Sensorimotor History Questionnaire (KSHQ)	Carrasco (1990) (48)	**NR**	**-**	**NR**	**NR**	**NR**	**NR**	**NR**	**NR**
Participation and Sensory environment Questionnaire– Home Scale (PSEQ-H Scale)	Pfeiffer (2018) (55)	NR	+	NR	-	NR	NR	NR	+
	Bevans (2020) (56)	+	+	NR	-	+	+	+	+
PSEQ: Home Scale: Overall Rating from all studies		**+**	**+**	**NR**	**-**	**+**	**+**	**+**	**+**
Participation and Sensory environment Questionnaire– Community Scale (PSEQ-C Scale)	Pfeiffer (2019) (57)	**NR**	**+**	**NR**	**-**	**NR**	**NR**	**NR**	**+**
Sensory Behavior Questionnaire (SBQ)	Neil (2014) (58)	**NR**	**+**	**NR**	**NR**	**NR**	**+**	**+**	**+**
Sensory Experiences Questionnaire version 3 (SEQ-3.0)	Ausderau (2014) (49)	**+**	**NR**	**NR**	**NR**	**NR**	**NR**	**NR**	**+**
Sensory Processing Measure (SPM)	Parham (2007) (42)	-	+	NR	?	+	+	+	+
	Dugas (2018) (72)	NR	NR	NR	NR	NR	NR	?	NR
	Brown (2010a) (73)	NR	NR	NR	NR	NR	NR	?	NR
	Brown (2010b)(71)	NR	+	NR	-	NR	NR	?	NR
	Lai (2011) (69)	NR	NR	?	NR	NR	NR	NR	NR
	Ahmad (2020) (60)	NR	NR	+	NR	NR	NR	NR	NR
SPM: Overall Rating from all studies		**-**	**+**	**±**	**?**	**+**	**+**	**±**	**+**
Sensory Processing Measure- Preschool (SPM-P)	Parham (2010) (51)	-	+	NR	?	+	-	+	+
	Alkhalifah (2019) (70)	NR	NR	-	NR	NR	NR	NR	NR
SPM-P: Overall Rating from all studies		**-**	**+**	**-**	**?**	**+**	**-**	**+**	**+**
Sensory Processing Scale Inventory (SP Scale Inventory)	Schoen (2017) (52)	**+**	**+**	**NR**	**NR**	**NR**	**NR**	**NR**	**?**
Sensory Processing and self-regulation checklist (SPSRC)	Lai (2019) (50)	**?**	**+**	**NR**	**+**	**NR**	**NR**	**+**	**+**
Short Sensory Profile 2 (SSP2)	Dunn (2014) (41)	NR*	+	NR	+*	+*	+	+*	+*
	Chojnika (2019) (53)	NR	NR	+	NR	NR	NR	NR	NR
SSP2: Overall Rating from all studies		**NR**	**+**	**+**	**+**	**+**	**+**	**+**	**+**

Table S10: Quality of measurement properties per study based on COSMIN quality criteria

Standards for evaluating the quality of Measurement development	Assessment of Sensory Processing and Executive Functions in Childhood (EPYFEI) (Romero-Ayuso 2018) (47)	Participation and Sensory Environment Questionnaire (PSEQ) (Pfeiffer 2018) (54)	Child Sensory Profile 2 (CSP2) (Dunn 2014) (41)
Total Quality of the Measure Design	I	A	I
Total quality of Pilot study *	I	A	I
Total rating for Quality of measure development	**I**	**A**	**I**

Phase 1- Interview guide: Parent/ Carer semi-structured interview

The researcher will thank study participant(s) for coming and explain the purpose of research, interest in the participant(s), confidentiality, and recording, answer any questions before commencing, ensure consent forms are signed, and study participants have a copy.

''We value your insights and experiences as to what we as health professionals need to know about tic disorders. We have invited you here today as you have some personal experiences of either having a tic disorder or having a loved one with tics. Through understanding your or your child or young person's experiences with tics, we aim to develop a new assessment questionnaire that will help provide clear insights into the sensory areas that treatment needs to focus. The assessment tool aims to provide health professional with better insights into the sensory challenges that people with tics may be experiencing. As it may be common to have other existing conditions with tic disorders such as concentration difficulties, anxiety, sensory sensitivities or emotional dysregulation we want to focus on all these areas too and not just tics.''

Semi-structured interview Questions for study participants:

1. What do you want health professionals to know about your child or young person's experiences with tic disorders?

2. Has your child / young person experienced any of these sensory sensitivities to touch, taste, sight, hearing, smell, movement, our body awareness or to temperatures, and if so, what are these? (Use sensory poster/ prompt to facilitate discussion- see appendix 10)

3. How do these sensory sensitivities impact your child's or young person's participation in daily life activities? (Use activities of daily living poster/ prompt to facilitate discussion- see appendix 8)

4. Are there things that you feel are important that we need to ask children or young people with tics to better understand what they are going through?

5. If emotional regulation is difficult for your child or young person with tics, how does it impact on their life or engagement in activities?

6. Which environments are the most challenging for your child / young person to deal with (i.e. home, school, school assembly, school camps, shops, sports events, etc.) and why?

7. What issues or conditions or symptoms have had an influence on your child or young person with tics ability to participate in daily tasks?

8. What do you want your child or young person's health team to focus on when treating them?

9. What are the strengths or enablers your child or young person has experienced due to tics or other existing conditions that have supported you or them to engage in activities or participate?

10. Is there anything else that you think might be important to mention?

Phase 1- Interview guide: Young person semi-structured interview

The researcher will thank the study participant(s) for coming and explain the purpose of research, interest in the participant(s), confidentiality, and recording, answer any questions before commencing, ensure consent forms are signed, and study participants have a copy.

''We value your insights and experiences regarding what we as health professionals need to know about tic disorders. We have invited you here today as you have some personal experiences with tics. Through understanding your experiences with tics, we aim to develop a new assessment questionnaire that will help us know what questions we need to out in the new questionnaire. The new questionnaire aims to provide health professionals with better insights into all the difficulties that people with tics may be experiencing. As it may be common to have other existing conditions with tics, such as concentration difficulties, anxiety, sensory sensitivities or emotional dysregulation, we want to focus on all these areas too and not just tics, if that is Ok with you?

Semi-structured interview Questions for study participants:

1. What do you want health professionals to know about your experiences with tic disorders?

2. Have you experienced any of these sensory sensitivities to touch, taste, sight, hearing, smell, movement, body awareness, or temperatures, and if so, what are these? (Use sensory poster/ prompt to facilitate discussion- see appendixes)

3. How do these sensory sensitivities impact your participation in daily life activities? (Use activities of daily living poster/ prompt to facilitate discussion- see appendixes)

4. Are there things you feel are important we need to ask children or young people with tics to understand better what they are going through during assessment and planning treatment?

5. Is managing your emotions something tricky for you?
   If so, how does it impact your life or you do things?

6. Which environments are the most challenging for you to deal with (i.e. home, school, school assemblies, school camps, shops, sports events, etc.) and why?

7. What difficulties have you experienced that have influenced your ability to participate in daily tasks?

8. What do you want your health team to focus on when treating you?

9. What strengths do you have that have supported you to engage in activities?

10. Is there anything else that you think might be important to mention?

This document provided a detailed description of the data analysis process of this qualitative study. The researchers ensured the study's trustworthiness by being credible, transferable, dependable and confirmable [60] by implementing the six phases of thematic analysis, an iterative and reflective process, to analyse the data [54, 55, 61, 62]. These phases were: familiarising yourself with the data (Phase 1), generating initial codes (Phase 2), searching for themes (Phase 3), reviewing the themes (Phase 4), defining and naming the themes (Phase 5) and producing the report (Phase 6) (Figure 1)[55].

Phase 1**: **Familiarise yourself with your data

All interviews were transcribed verbatim and proof checked against the audio-video recordings. This allowed the transcripts to be read several times before initial codes were generated and for the researchers to have prolonged engagement with the data whilst searching for meaning and patterns. The raw data was stored in well-organised archives, such as NVivo 12 software [59] and Excel spreadsheets for demographic data. All study participants were provided with pseudonyms to protect their privacy and identity. Memos were kept concerning reflections on the data using NVivo 12 Software. Team members read transcripts, and meetings were held to triangulate and discuss different potential codes and themes. All thoughts, impressions and informative points were recorded in the principal researcher's journal.

Phase 2**: **Generating initial codes

Fortnightly meetings with the principal investigator (NS) and research supervisor (PB) were undertaken regarding information gained from semi-structured interviews and to discuss findings in assisting with peer debriefing. Frequent meetings with the research team were held to examine and define codes, aiding researcher triangulation. Through

discussion and agreement from the research team, a coding framework, identifying codes and definitions of these codes, was developed. Copies of transcripts, journals, memos, email correspondence between the research team members, meeting agendas and meeting minutes were stored to ensure an audit trail of the generation of the codes. Hierarchical coding was used to allow the researchers to analyse the texts at varying levels of specificity, with higher-order codes providing an overview and lower-order codes providing more specific and detailed distinctions between study participants [54].

Phase 3: Searching for themes

An in-depth analysis of hierarchies of the different codes, concepts and themes was undertaken, including diagrams to make sense of the theme connections and to ensure a consistent approach was used for which detailed notes were taken [55]. This technique reduced the initial codes and themes by identifying similarities and grouping common codes. When identifying themes across the data sets, the researchers worked systematically through the entire data set, giving full and equal attention to each data item [54].

Phase 4: Reviewing the themes

Research team members vetted themes and subthemes by testing for referential adequacy by referring back to the raw data and the study participants' quotes [55].

Phase 5: Defining and naming the themes

Team consensus on the themes and the naming of these themes was achieved following researcher triangulation, peer debriefing and team meetings. Documentation of the team meetings regarding the agreement of themes and the naming of themes was kept [55].

Phase 6**: **Producing the report.

Credibility, which addresses the ‘‘fit’’ between study participants' experiences and the researchers' representation of them [63], was addressed in this study through prolonged engagement with the data, persistent observation, researcher triangulation, peer debriefing and member checking.

22 /06 /2022

Dear Study Participant
(Details to be inserted)

Dear,

Re: Preliminary findings from the research study you kindly participated in

Thank you and your family for your time and the experiences you shared with us through your involvement in a research study titled: **Development and trial of a sensory assessment measure for children & young people with tic disorders.** This research study was conducted by researchers at **The Children's Hospital at Westmead**. We hoped to clearly understand the sensory challenges children and young people with tics may be experiencing.

This letter serves multiple purposes. We want to:

- Thank you for your involvement in the study. Without your participation, we would not have been able to gain helpful insights into the sensory challenges that children & adolescents experience daily.
- Share the study's preliminary findings with you.
- Invite you to provide feedback on the findings to ensure that we have understood and interpreted your words and experiences correctly.

Results:

We had 16 families participate in interviews that ranged between 45 minutes and 2 hours in duration. Through analysing the information and experiences you shared with us, we were able to find common themes.

We identified that sensory struggles affect children & young people in all environments. We then divided these tasks into activities completed at home and in the community. Attached is a diagram to assist with explaining these themes and findings.

Environment: In the home environment
Theme: 1. We sacrifice and adapt to get daily activities done in the home

Children/ young people have difficulty dressing due to the feeling and texture of the clothes, impacting the type of clothes bought. Many children / young people had to choose the purchased clothes, or they would not wear them if they did not feel right. It was evident that it was a struggle

to get children/ young people to wear clothes, to wear the appropriate clothing for the weather, and many children/ young people preferred to wear shorts and a T-shirt, even in cold weather. Many Parents explained the different adaptions made to their child / young person's school uniform.

Mealtimes were very stressful as many families could not eat meals together. The main reason being that the chewing and slurping sounds made by other family members at the table were intolerable to their child / young person. The range of food eaten by the child / young person was explained to be very limited due to the texture, taste and smell of food.

Hygiene and grooming tasks were overwhelming due to the different sensory experiences involved in these activities (such as the feeling of nails or hair being cut or the taste of the toothpaste). Particular brands or products were purchased to assist in making these activities easier for children/ young people.

A good sleep routine was a struggle as the sheets, and the weight of the blankets had to feel right. Many children / young people preferred not to wear clothing in bed, which made it challenging for parents to ensure that their children were warm through the night.

Due to the environmental temperature, mainly if it was hot, children had difficulty with increased emotional distress, and in some cases, their tics were noted to be worse. Children experiencing emotional distress due to the TV or radio being too loud was a common challenge.

Environment: In the community
Theme: 2. Environments hinder my child's participation in the community

Parents bought online to avoid taking their child / young person to the shops due to the sensory experiences, i.e. loud noises and crowding, which caused emotional distress. The smell of public toilets and the sounds (toilets flushing and the hand dryers) meant avoiding using these amenities. Travelling in a car or public transport due to the proximity to other people, the sounds these people made, and the radio meant it is challenging to travel places.

Children identified that the school environment was very loud and challenging due to the ongoing bombardment of sensory input all day. Attending events such as a fireworks display, school disco or party or even eating out at a restaurant could cause significant emotional distress.Therefore, many parents explained that they stayed at home rather than attend events.

Theme: 3. These problems impact our child and family

As a result of the sensory dysregulation the child or young person experienced, there was an impact on any additional conditions, such as tics and increased anger or emotional distress, affecting the

entire family. Families lived life in a predictable way, such as always buying clothing items from the same store, cutting out tags, making additional meals for their child/ young person in place of the family meals etc., to keep the peace and ensure their child / young person could cope. Many families mentioned that the anger and emotional dysregulation experienced were worse than any of the other symptoms or conditions, and in some cases, children may harm other family members. Young people also explained that when they became emotionally distressed from the sensory input, there was an increase in the tics that they experienced. This then would make them even more upset.

Proving your feedback:

We would be most grateful if you would provide us with feedback on the preliminary results and our understanding of the problems children / young people experience to ensure we have interpreted your words and experiences correctly.

Please answer the questions provided and send them back to the researcher via the email below. Should you prefer to answer these same questions over the phone with the researcher, please email times that are convenient for you and we can arrange a date and time to call you.

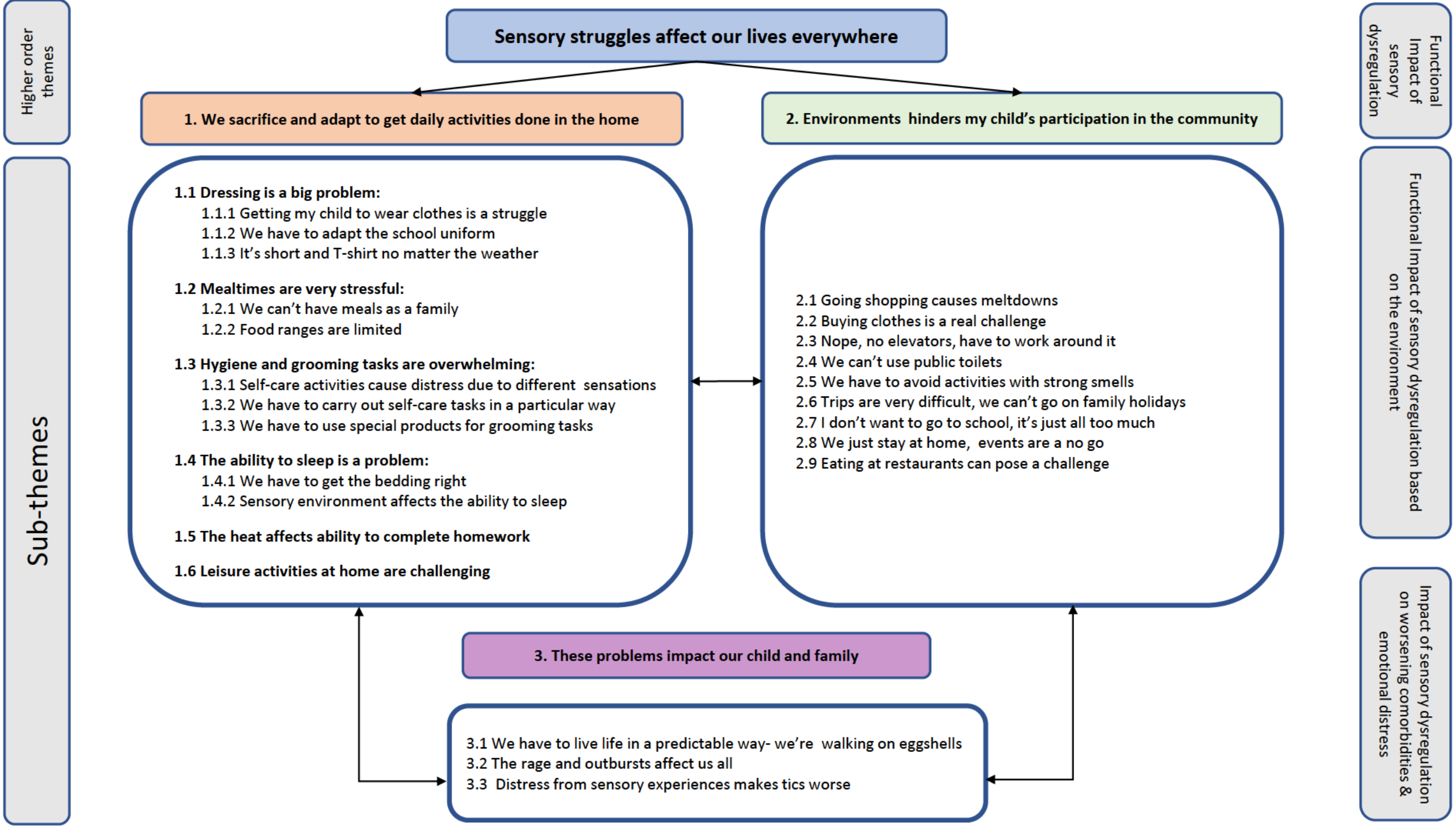
Participation in providing feedback on the findings in this research study is voluntary. If you do not wish to participate, you do not have to. You and your child / your young person will receive the best possible care whether or not you take part.

The researcher's contact details for this study are: **Nicolette Soler: (02) 9845-2005 or email address: nicolette.soler@health.nsw.gov.au or**

Please do not hesitate to contact us if you require any further information or wish to discuss this study in more detail.

Yours sincerely,

Nicolette Soler
Principal Investigator
Department of Paediatric Neurology& Psychological Medicine, The Children's Hospital at Westmead


Higher order themes
Sub-themes
Sensory struggles affect our lives everywhere
1. We sacrifice and adapt to get daily activities done in the home
2. Environments hinders my child's participation in the community
1.1 Dressing is a big problem:
1.1.1 Getting my child to wear clothes is a struggle
1.1.2 We have to adapt the school uniform
1.1.3 It's short and T-shirt no matter the weather
1.2 Mealtimes are very stressful:
1.2.1 We can't have meals as a family
1.2.2 Food ranges are limited
1.3 Hygiene and grooming tasks are overwhelming:
1.3.1 Self-care activities cause distress due to different sensations
1.3.2 We have to carry out self-care tasks in a particular way
1.3.3 We have to use special products for grooming tasks
1.4 The ability to sleep is a problem:
1.4.1 We have to get the bedding right
1.4.2 Sensory environment affects the ability to sleep
1.5 The heat affects ability to complete homework
1.6 Leisure activities at home are challenging
2.1 Going shopping causes meltdowns
2.2 Buying clothes is a real challenge
2.3 Nope, no elevators, have to work around it
2.4 We can't use public toilets
2.5 We have to avoid activities with strong smells
2.6 Trips are very difficult, we can't go on family holidays
2.7 I don't want to go to school, it's just all too much
2.8 We just stay at home, events are a no go
2.9 Eating at restaurants can pose a challenge
3. These problems impact our child and family
3.1 We have to live life in a predictable way- we're walking on eggshells
3.2 The rage and outbursts affect us all
3.3 Distress from sensory experiences makes tics worse
Functional Impact of sensory dysregulation
Functional Impact of sensory dysregulation based on the environment
Impact of sensory dysregulation on worsening comorbidities & emotional distress

Parent's Name: _____ (Optional)

Child / young person's name: _____ (Optional)

Thank you so very much for completing these questions. By gaining your feedback from our preliminary results, ensure that we have interpreted your words and experiences correctly. The questionnaire should take 5-10 minutes of your time to complete.

Once completed, please email this questionnaire to the principal researcher, Nicolette Soler, at Nicolette.soler@nsw.gov.au

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Please circle the response below that best fits your experience.

These three questions will relate to the first theme:

1. We sacrifice and adapt to get daily activities done in the home

	Response
1. Do to feel the wording used for this them describes what you and your child experience? If not, what wording would you suggest to describe your experiences best? _____	**Yes / No**
2. Do you feel the results reflect all your responses and experiences your child / young person has regarding sensory struggles impacting their life at home? If not, what should be included or excluded:	**Yes / No**
3. Do you think this theme makes sense to you? If not, please explain:	**Yes / No**

These three questions will relate to the second theme:

2. Environments hinder my child's participation in the community

	Response
4. Do to feel the wording used for this them describes what you and your child experience? If not, what wording would you suggest to describe your experiences best? _____	**Yes / No**
5. Do you feel the results reflect all your responses and experiences your child / young person has regarding sensory struggles impacting their life at home? If not, what should be included or excluded:	**Yes / No**
6. Do you think this theme makes sense to you? If not, please explain:	**Yes / No**

These three questions will relate to the third theme:

3. These problems impact our child and family

	Response
7. Do to feel the wording used for this them describes what you and your child experience? If not, what wording would you suggest to describe your experiences best? _____	**Yes / No**
8. Do you feel the results reflect all your responses and experiences your child / young person has regarding sensory struggles impacting their life at home? If not, what should be included or excluded:_____	**Yes / No**
9. Do you think this theme makes sense to you? If not, please explain:_____	**Yes / No**

10. Overall, do you think this model with the three themes makes sense to you? **Yes / No**
    If not, please explain:_____

_____

Is there any additional feedback or comments you would like to make?_____

_____
_____
_____
_____

Thank you so very much for your time and support of our research. We will provide you with the final results once we compile the feedback and make the necessary changes.

Please do not hesitate to contact us if you require any further information or wish to discuss this study in more detail.

Yours sincerely,

Nicolette Soler
Principal Investigator
Department of Paediatric Neurology& Psychological Medicine,
The Children's Hospital at Westmead