Communication in Children with Tourette Syndrome

by

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Abstract

Tourette syndrome (TS) is a neurodevelopmental condition involving motor and vocal tics that begins in childhood. Children with TS often present with other co-occurring conditions such as Attention-Deficit/Hyperactivity Disorder and some have challenges in communication development. The overall purpose of this research was to improve understanding of communication development in children with TS. The aims of the research were to collate and summarize past research, measure communication skills, explore how psychosocial skills and co-occurring conditions relate to communication skills, and describe communication in day-to-day contexts. Three studies were conducted to address these aims.

The first study was a scoping review that maps, describes, and summarizes the literature reporting on the language and social communication skills of children with TS and identifies knowledge gaps. It is described in Chapter 2. A systematic search of six electronic databases revealed 56 studies that measured language or social communication. The majority of identified studies measured verbal IQ or verbal fluency. This literature suggests that children with TS have strengths in verbal intelligence, story/sentence recall, categorisation, and performance on tasks at the single-word level. Moreover, a subset of children with TS had language disorders and social communication challenges, with rates of language delay/disorder being higher in children with co-occurring conditions (i.e., 20-45%). There was little research investigating language beyond the single-word level as well as a lack of detailed reporting about the nature of language and social communication challenges in children with TS.

Using a series of parent questionnaires to measure communication skills and psychosocial skills, the second study describes the proportion of children in a North American sample of children with TS who present with challenges in communication development,

compares communication skills to a normative sample, analyzes how co-occurring Attention-Deficit/Hyperactivity Disorder, Obsessive Compulsive Disorder, and Anxiety relate to communication skills, and investigates if communication skills correlate with psychosocial skills. This study is described in Chapter 3. Information was collected using the Children's Communication Checklist, Second Edition and the Strengths and Difficulties Questionnaire. Sixty-two percent of children received scores indicating age-expected communication skills and 38% obtained scores suggesting communication challenges. Mean scores for the children with TS were significantly below age-expected norms for general communication and social-pragmatic composites, as well as for multiple domain subscales. The presence of co-occurring conditions did not predict communication challenges. There was a negative correlation between communication scores and psychosocial scores: when communication scores decreased, psychosocial challenges increased.

The third study was a scaffolded mixed methods case series that aimed to describe language development and day-to-day communication in five children with complex TS (i.e., TS with other co-occurring conditions). This study is described in Chapter 5. The study reports language scores across core language, narrative language, high-level language (e.g., inferencing), and social communication domains of language development. Communication log books and semi-structured parent interviews were used to understand day-to-day communication. School accommodations, extra services, or alternative education options were being accessed by many parents. Parents used proactive approaches to manage day-to-day communication in the home and they strategized to keep the environment calm and productive. Parents also adopted attitudes of acceptance and understanding. In some cases, parents provided social coaching and created intentional social experiences so that their children could be successful.

The combined findings of this research indicate that many children with TS will develop age-appropriate language skills; however, many others may experience challenges across a range of language and social communication areas. The second and third studies provide evidence that young children with TS are at risk of having their communication challenges overlooked. For children referred to a speech-language pathologist, assessment should go beyond investigating core language skills, considering narrative language, high-level language, and social communication. I also recommend collecting background information about co-occurring conditions, the severity of the tic disorder, and levels of psychosocial functioning. Treatment approaches should consider the child's psychosocial and emotional-behavioural needs. Creating a calm environment can help a child manage anxiety and other tic triggers. A flexible approach can help children participate in traditional forms of intervention. The SLP can also have a role in shaping positive social environments for children by educating others about TS and promoting acceptance of the social differences that go along with this neurological difference.

Preface

This dissertation represents original work completed by Angela Feehan under the supervision of Dr. Monique Charest, Associate Professor, Department of Communication Sciences and Disorders. The supervisory committee provided guidance throughout the work: Drs. Paola Colozzo, Lesley Pritchard, and Veronica Smith.

Chapter two of this thesis has been published as:

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I took on the primary role in completing this scoping study, including planning the search, review of records, data extraction, compiling the narrative summary, and writing the manuscript.

Chapter three of this thesis will be revised for submission to a peer-reviewed journal. The manuscripts will be co-authored with Drs. Charest, Colozzo, Pritchard, and Smith. I planned, compiled and disseminated the survey, analyzed data, and wrote the manuscript all with guidance from the co-authors.

Chapter four of this thesis will be revised for submission to a peer-reviewed journal. The manuscripts will be co-authored with Drs. Charest, Colozzo, Pritchard, and Smith. I planned the study, recruited participants, conducted assessments and interviews, analyzed data, and wrote the manuscript, all with guidance of the co-authors.

The research projects in chapters three and four were approved by the University of Alberta Health Research Ethics Board:

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Dedication

This work is dedicated to everyone living with TS. May you talk and listen in ways that bring you lasting joy.

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Abbreviations

ADHD – Attention-Deficit/ Hyperactivity Disorder

CASL-2 – Comprehensive Assessment of Spoken Language, Second Edition

CCC – Children's Communication Checklist, First Edition

CCC-2 – Children's Communication Checklist, Second Edition

CELF-5 – Clinical Evaluation of Language Fundamentals, Fifth Edition

C-TS / Complex TS – Tourette syndrome with other co-occurring conditions

GTRS – Global Tic Rating Scale

OCD – Obsessive-Compulsive Disorder

Pure TS – TS without other co-occurring conditions

SDQ – Strengths and Difficulties Questionnaire

TNL-2 – Test of Narrative Language, Second Edition

SLP/SLPs Speech-language pathologist(s)

SRS – Social Responsiveness Scale

TS – Tourette syndrome

Chapter 1: Introduction to Communication in Children with Tourette Syndrome

Although communication has not traditionally been viewed as a core concern in Tourette syndrome (TS), individuals with TS are known to perform differently on executive functioning and social information processing tasks. Some of these tasks represent skills that are essential in communication. Recently, researchers have explored the hypothesis that social communication challenges are a central component of TS. However, there has been little research regarding communicative functioning in the TS population to date and limited research with children, in particular. Understanding communication in childhood is of particular interest as it is the period when communication skills are emerging. A review of the literature related to communication among individuals with TS indicates several research needs: There is a need to understand to what extent children with TS experience challenges with communication functioning, the nature of communication challenges, which children experience these challenges within the broad spectrum of TS, how communication skills are related to overall levels of functioning, and what other factors may be important in relation to communication functioning (e.g., communication environment).

This thesis addresses the aforementioned gaps in the research on communication in children with TS. In this introductory chapter, I will provide a working definition of communication and describe the communication theories being used across studies. I will also provide an introduction to TS, describe literature on the cognitive aspects of TS (social cognitive skills and neuropsychological skills) and summarize studies that have measured communication skills in children with TS. Next, I will present the purpose of the dissertation, providing an overview of the three studies that were conducted to address seven research questions. Finally, I

will give a detailed background about the methodology and methods, interpretive frameworks, and theoretical perspectives used in the research to demonstrate how methodological coherence has been maintained in the mixed methods study (Chapter 5).

The first study presented in this dissertation was a scoping review of literature on language and social communication in children with TS and is described in Chapter 2. The second study was a parent-reported measures study of communication and psychosocial skills and is described in Chapter 3. Additional psychometric information from the second study is described in Chapter 4. Chapter 5 describes the third study, a series of mixed methods case studies about the communication skills of five children with TS. Additional methodological information from the third study is described in Chapter 6. Chapter 7 is a general discussion of the findings of this research.

Introduction to Communication

This section will describe how communication is being considered across studies in this dissertation. Communication is defined as *the sending and receiving of communicative intent between partners through verbal and nonverbal actions.* Huang (2015) proposed two broad ways of viewing communication (presented in Hyter, 2017). The first, the Anglo-American view, posits that communication is the "use" of the language system, which is made up of specific domains (e.g., phonology, morphosyntax, and semantics). The second view, the Continental (European) view, emphasizes how language meaning and use are indistinguishable from cognitive, social, and cultural contexts. The Anglo-American and Continental views are not mutually exclusive, and in fact, Hyter (2017) takes the position that incorporating both can provide a more comprehensive perspective on communication. Others (such as Barnes & Bloch, 2019) suggest that there has been too much focus on the Anglo-American way of viewing

communication and that we should shift our focus toward the real-life contexts in which communication occurs (in line with the Continental view). Barnes & Bloch emphasize that communication is a phenomenon occurring in the co-presence of individuals and in real-time, not in the minds of any single individual.

This dissertation has considered both approaches to bring these two important viewpoints together and begin expanding research that takes a Continental viewpoint. The Bloom & Lahey (1978) *form*, *content*, and *use* model is being used in this research to organize the Anglo-American viewpoint. Language *form* refers to morphology and syntax, language *content* refers to semantics/meaning, and language *use* refers to social communication. To expand on the foundational work of Bloom and Lahey, it is important to note that several contemporary theories also include aspects of cognition as building blocks for communication, including social cognition, attention, memory, executive function, and affective skills (e.g., Hyter, 2017; Wiseman-Hakes, 2021). For example, attention skills are needed to detect social cues and maintain an interaction, memory is needed to follow a conversation, executive functions regulate specific skills like the planning of messages, and affective regulation skills maintain emotional states conducive to communication. It is important to consider these cognitive building blocks of communication in order to understand some of the ways in which children with TS may experience disruption to their communication experiences. To distil the Continental viewpoint of communication, *functionalism* (Bates, 1991) was the theory used. It is described in detail later in this chapter in a fulsome description of the philosophical and theoretical positions that guided the design and analysis in Chapter 5.

Chapter 2 covers literature from an Anglo-American viewpoint (as no research from a Continental perspective currently exists) and Chapter 3 also takes an Anglo-American viewpoint

through the use of a pre-developed communication assessment tool that fits within the Anglo-American tradition. Chapter 5 incorporates tools from the Anglo-American tradition, thereby grounding readers with this familiar perspective, but it also integrates the Continental approach by considering communication in real-life contexts and day-to-day interactions. The Bioecological model was employed to further explain how these contexts are being considered. A description of this model and its application in this research is described later in this chapter along with functionalism.

In Chapter 2, areas of communication are considered under the terms 'language' (i.e., form and content) and 'social communication' (i.e., use). For the purposes of Chapter 2, ' language' encompasses expressive and receptive language skills, considering semantics, morphosyntax, narrative language, and supralinguistics (i.e., high-level language). To define 'social communication,' Chapter 2 uses the Adams (2005) framework, which includes *social interaction*, *social cognition,* and *pragmatics* (verbal and nonverbal). Adams' framework for social communication also includes *language processing* (expressive and receptive language); however, for the purposes of Chapter 2, language processing falls within the 'language' domain. Additionally, since some aspects of autistic traits are based on social communication, autistic traits were also organized under social communication in Chapter 2.

In Chapter 3, the terms 'communication' and 'general communication' are used to refer to language and social communication skills combined (form, content, *and* use). Variables in Chapter 3 were defined based on the constructs of the Children's Communication Checklist, Second Edition (CCC-2; Bishop, 2003). The CCC-2 provides a General Communication Composite that includes *speech*, *syntax*, *semantics*, *coherence*, *initiation*, *scripted language*, *context*, and *nonverbal communication*, thereby covering aspects of form, content, and use. The

term ‘social-pragmatic communication’ and the Social-Pragmatic Communication Composite refer to social communication only (i.e., use). This composite includes *initiation*, *context*, and *nonverbal communication* domains of the CCC-2 and was chosen to parallel the research of Saul et al. (2022). An additional domain of social communication that the CCC-2 investigates (but that is not included in the Social-Pragmatic Communication Composite), is *scripted language*. Chapter 3 also investigates two domains that describe autistic traits, *social relations* and *interests*.

As previously mentioned, Chapter 5 incorporates both Anglo-American and Continental viewpoints of communication. As described above, the CCC-2 domains were employed under the Anglo-American tradition (as in Chapter 3). Similarly, additional communication assessment tools were used to investigate core language, narrative language, and high-level language domains. The Continental perspective was incorporated in Chapter 5 by investigating parent reports of day-to-day communication experiences in different real-life contexts. A mixed methods approach (whereby Anglo-American-based communication information was collected using quantitative tools and Continental-based communication information was collected using qualitative tools) was ideal for combining the two traditions as it provided an established methodological approach for mixing and combining two very different types of data.

Introduction to Tourette Syndrome

TS is a neurodevelopmental condition characterized by motor and vocal tics that begin in childhood (American Psychological Association, 2013). TS occurs in about 0.3-0.9% of the population (Scharf et al., 2015), and boys are affected more than girls by a factor of 3.6 (Scharf et al., 2012). Although little is known about genetic correlates in TS, it has been established as a highly heritable condition (Albin, 2018). Several neuroanatomical differences have been

connected to TS; the most well-supported are differences in the basal ganglia, specifically striatal differences (Albin, 2018). The typical course of TS is that the severity of tics increases in early adolescence and then wanes in adulthood (Martino & Leckman, 2013). In the majority of cases, TS is accompanied by co-occurring conditions including Attention-Deficit/Hyperactivity Disorder (ADHD), Obsessive-Compulsive Disorder (OCD), and Anxiety (70-90% of cases; Cath et al., 2022). Individuals who have co-occurring conditions are sometimes referred to as "complex," (vs. "pure" TS, TS without additional diagnoses) as the addition of other diagnoses increases the complexity of the clinical presentation.

TS Beyond Motor Symptoms

Although TS is a diagnosis defined by motor and verbal tics, there are several findings in the literature to suggest that further research into communication development and social experiences in this population is warranted. In particular, several recent proposals have posited that aspects of social communication may be more central to TS than previously considered (Albin, 2018; Eddy, 2021). To contextualize this recent interest in social communication and to establish future research directions, I will present background information about social cognition, neuropsychological skills, language development, the features of co-occurring conditions, social communication, and social/psychosocial functioning.

Social Cognition

Social cognition is thought to be an important building block of communication (Hyter, 2017; Wiseman-Hakes, 2021). Thus, social cognitive differences can be expected to be evident in communication for individuals with TS. In recent years, adults with TS have been found to experience specific social cognitive differences that presumably affect how they interact in the social world, including problems with comprehending and interpreting the intentions of others,

recognizing/judging social indiscretions, perspective-taking, and regulating emotions during stressful interpersonal interactions (Channon et al., 2012; Eddy et al, 2010; Eddy & Cavanna, 2013; Eddy et al., 2015). Conversely, some studies have found that adults with TS have similar social cognitive skills compared to nondisabled adults (i.e., theory of mind, empathy, identifying mental state from facial expression; Channon et al., 2004; Baron-Cohen et al., 1997). These inconsistent findings may be due to differences in tasks. Specifically, Eddy (2021) suggests that social cognitive challenges may be nuanced in individuals with TS and that they may emerge when task complexity increases. Literature investigating social cognition in children with TS is sparse and, to date, findings have been mixed. Children with TS have demonstrated lower overall social cognition scores measured with a parent survey (Güler et al., 2015) and challenges understanding indirect sarcasm (Drury et al., 2018). For those who also had ADHD, recognizing angry prosody was a challenge when it was incongruent with the content of the message presented (e.g., 'I'm so excited for my birthday' being said in an angry tone of voice; Drury et al., 2012).

In contrast, children with TS have performed similarly to nondisabled peers when answering questions about mental states, understanding direct sarcasm, and discriminating and naming emotions on faces (Drury et al., 2018; Drury et al., 2012). The specific tasks completed in research with children have differed from those completed with adults and the child literature is scant, making comparison difficult. If differences exist between adults and children, they may be due to changes in the TS population as they age. That is, many young people experience a decrease in tics after adolescence accompanied by a removal of the TS diagnosis. Those who persist with having TS into adulthood may represent a different group with more significant challenges.

Neuropsychological Profiles

Compared to nondisabled peers, children with TS have been found to have challenges in neuropsychological domains, including divided and sustained attention (Chang et al., 2007; Sherman et al., 1998), long-term nonverbal memory (e.g., Bloch et al., 2006), and executive control (Sikora et al., 2019). These neuropsychological differences could lead children with TS to experience challenges in everyday communication contexts. In particular, attention, memory, and executive function are included in models of communication and are thought to provide a foundation for development and use (Hyter, 2017; Wiseman-Hakes, 2021).

Oral Language Development in Children with TS

Language skills (encompassing language form and content) such as vocabulary, sentence structure, and grammar, are essential to successful communication. We can gain insight into the oral language skills of children with TS by looking at studies that have reported language skills and disorders among children with TS.

Prevalence of language disorders. Several studies have found that children with complex TS (TS with other co-occurring disorders) have a higher prevalence of language disorders than would be expected in the general population. A population-based study of children with TS found that 30% presented with a language disorder according to parents' reports (Claussen et al., 2018). Language delay/disorder incidence is estimated at 3-6% for children with TS who do not have co-occurring conditions (representing about 20% of the TS population) and 20-45% for children with TS who do have co-occurring conditions (representing about 80% of the TS population; Cravedi et al., 2018; Spencer et al., 1998; Cath et al., 2022). While these studies suggest that children with complex TS can experience co-occurring language disorders,

previous research provides limited insight into the language profiles (the specific areas of relative strength and weakness) seen in language disorders associated with TS.

Language Skills. Research on the language skills of children with TS has largely focused on tasks at the single-word level and there has been a great deal of variability in the tasks used to investigate language skills. There has also been some research looking at verbal intelligence, expressive language fluency, narrative language, high-level language (e.g., inferencing and non-literal language), and a few studies measuring aspects of expressive and receptive language. The findings of this research can be organized into studies that indicate language strengths and studies that indicate potential language challenges.

Language skills – Strengths. Many studies have demonstrated age-expected vocabulary, verbal intelligence, sentence repetition, and verbal fluency (word production) in children with TS (e.g., Carter et al., 2000; Church et al., 2009; De Groot et al., 1997; Hulbert, 1986; Jensen, 2004). Moreover, some research has documented that children with TS produce responses as accurately and faster than nondisabled peers for certain single-word expressive tasks (Dye et al., 2016; Walenski et al., 2007).

Language skills – Potential Challenges. Children with TS have been found to have challenges with fluent expressive language formulation when speaking in sentences. While they do not generally demonstrate a greater number of stuttering-like dysfluencies, they produce a greater number of typical disfluencies (disfluencies related to language formulation) compared to nondisabled peers, such as a greater number of partial word repetitions, phrase repetitions, and sentence revisions (de Nil et al., 2005; Donaher, 2008). De Nil et al (2005) found that children with TS produced a similar number of formulation disfluencies regardless of whether or not they

had other co-occurring conditions. Expressive fluency challenges may negatively impact communication success in children with TS.

Sukhodolsky et al. 2003 measured parent-reported communication using the Communication subscale of the Vineland Adaptive Behaviour Scale (Sparrow et al., 1984), which includes questions about receptive, expressive and written communication. Children with TS were rated by their parents. The children with pure TS did not differ significantly from a group of nondisabled peers; however, children with TS and ADHD had lower communication scores compared to nondisabled peers.

Using a series of cases, Legg et al. (2005) investigated narrative language and high-level language skills (e.g., inferencing and non-literal language) in 10 children with TS. Seven of the 10 children had challenges with at least one of the subtests of the Test of Language Competence, which assesses understanding ambiguity, inferencing, expressing intentions in sentences, and understanding metaphors (Wiig & Secord, 1989). Of those seven children, three had co-occurring ADHD, and six had co-occurring OCD. Five of 10 participants experienced challenges with the narrative fable task, including difficulty retelling the fable and difficulty producing a summary. Of those five, two had co-occurring ADHD and three had co-occurring OCD. Keeping in mind the small numbers in this sample, it's important to note that the groups with language needs included children with and without co-occurring conditions. Difficulties with more complex language tasks such as understanding non-literal language and story retelling could also be linked with neuropsychological and social cognitive differences that present in individuals with TS, since more complex language tasks tax memory and require skillful coordination of executive skills.

Language skills – Summary. In summary, although children with complex TS have higher rates of language disorders, studies examining performance on standardized language tasks for groups of children with TS have largely found age-expected scores in most areas of language. The literature indicates that challenges may lie in expressive formulation, high-level language, and narrative language and that some children with pure TS are also affected (i.e., de Nil et al., 2005; Legg et al., 2005); however, there is still very little research to draw from. Effective language formulation is crucial for communicating intent in real-time environments of everyday social interactions. High-level language is defined as, "the ability to use multiple areas of complex linguistic and cognitive processing" (Berg et al., 2002, p. 65). High-level language skills for tasks such as inferencing and non-literal language comprehension are necessary for social-communicative interactions, as these interactions require complex and efficient coordination of linguistic and cognitive processing (Lethlian & Murdoch, 1997). Narrative language is a form of discourse that also requires the coordination of multiple linguistic and cognitive skills (e.g., expressive and receptive language, memory, and executive function). Research has not yet provided descriptive information about how these complex and nuanced language challenges may manifest in day-to-day communication for children with TS. Several other factors relevant to communication in TS must be outlined to present a fulsome picture, as it can be currently understood. Similarly, there has not been research comprehensively assessing the communication system, including form, content and use and expressive/receptive language modalities. Next, I will turn to the communication features that characterize common co-occurring conditions.

Co-occurring Diagnoses in TS and Communication (Complex TS)

When studying communication in TS, it is important to consider the added complexity arising from the communication features associated with co-occurring conditions such as ADHD and OCD. Recall that these conditions are present in approximately 80% of people with TS (Cath et al., 2022). This section will describe the literature on co-occurring diagnoses with TS and what is known about them in relation to questions about communication. For each co-occurring diagnosis, I will discuss how this literature fits with the goal of studying communication in TS.

Attention-Deficit/ Hyperactivity Disorder (ADHD). ADHD is present in about 60% of individuals with TS (Martino & Leckman, 2013). ADHD in individuals with TS is related to greater challenges in working memory, visuomotor integration, inhibition, cognitive flexibility, and attention skills (Morand-Beaulieu, 2017). ADHD combined with TS has also been linked to problems with internalizing and externalizing, family function, psychosocial functioning, quality of life, and academics (Martino & Leckman, 2013).

Communication in ADHD. Certain items in the diagnostic criteria for ADHD (Diagnostic Statistical Manual, Fifth Edition; DSM-5; American Psychological Association, 2013) involve communication behaviors (e.g., does not listen when spoken to, talks excessively, talks out of turn). Individual and meta-analytic studies have established that aspects of communication are often a challenge in children with ADHD. Areas of difficulty identified were pragmatics (i.e., social communication), high-level language/discourse, and language formulation, whereas vocabulary and morphosyntax were within the expected range of skills (Helland et al., 2014; Korrel et al., 2017; Vaïsänen et al., 2014; Redmond et al., 2004).

Some studies have investigated language skills in groups of children with pure TS compared to those who had TS plus ADHD. Both social interaction and social communication

skills are a greater area of challenge for children with TS plus ADHD (Carter et al., 2000; Güler et al., 2015). The prevalence of language disorders is 20% in children with both conditions compared to only 6% in children with pure TS (Spencer et al, 1998). One study found that children with both TS and ADHD performed more poorly than children with pure TS on a categorization task that tested concept formation (i.e., semantics; De Groot et al., 1997). However, in the Legg et al. (2005) case series mentioned above, of 7 children with high-level and/or narrative language challenges, three had co-occurring ADHD and four did not. Additionally, studies have found that verbal intelligence scores in children with TS are similar regardless of whether or not they have ADHD (Carter et al., 2000; De Groot et al., 1997). Sukhodolsky et al. (2003) found similar parent ratings for children with TS for expressive, receptive, and written language regardless of ADHD co-occurrence (however, children with TS and ADHD had significantly poorer scores compared to nondisabled peers, but children with pure TS did not).

Overall, some studies indicate that co-occurring ADHD plays a role in communication functioning for children with TS. In particular, language disorders and problems in categorization, social communication, and social interaction appear to be more common when ADHD is present. On the other hand, verbal intelligence, high-level language, and narrative language skills have not been specifically attributed to ADHD co-occurrence in children with TS. Conclusions regarding the role of ADHD co-occurrence in expressive and receptive language ability are difficult to make based on the very limited current literature. Nonetheless, it seems reasonable to conclude that co-occurring ADHD should be considered carefully in future studies.

Obsessive-Compulsive Disorder (OCD). Reported rates of OCD vary from 10-35% in individuals with TS (Martino & Leckman, 2013) but reports of the occurrence of obsessive-compulsive behaviours (OCB) are much higher (60-90%; Martino & Leckman, 2013). OCD is thought to be closely related to TS as the two disorders cluster in families (Martino & Leckman, 2013). OCD is sometimes described as part of a TS "spectrum" (Albin, 2018, p. 333), or as a "nosological" relative of TS (Cravedi et al., 2018, p. 945). Eapen et al. (2016) reported that OCD co-occurrence is related to lower quality of life in childhood.

Communication in OCD. There are no communication-related diagnostic criteria for OCD and no known studies specifically linking challenges with language form or content to OCD. However, OCD may involve verbal and nonverbal behavior that is directed at managing stress associated with obsessions. Some researchers have reported that pragmatic language challenges occur commonly in the family members of those who have OCD (Cullen et al., 2008; Samuels et al., 2014). Another study suggested that adolescents and adults with OCD had greater challenges with nonverbal communication and other pragmatic language skills (unspecified by the authors) compared to nondisabled individuals (Ghahari et al., 2017), but no challenges were found in conversational skills, peer skills, or speech act use. Only a few studies have compared aspects of communication in children with TS who do and do not have OCD. Darrow et al. (2017) found that when children with TS also had OCD, they had stronger social communication scores on the Social Responsiveness Scale, Second Edition (SRS; Constantino & Gruber, 2012) compared to groups without a co-occurring OCD diagnosis. De Groot et al. (1997) found that children with TS performed more poorly on a categorization task that tested concept formation (i.e., semantics) and a measure of verbal intelligence when they had a co-occurring diagnosis of OCD. Given this information, a co-occurring diagnosis of OCD may contribute to some

communication challenges in children with TS, but the portrait of expected areas of strengths and weaknesses remains murky.

Depression and Anxiety. Rates of depression in individuals with TS range from 23-41% and rates of anxiety range from 16-55% (Martino & Leckman, 2013). Rates of depression and anxiety are higher in individuals with higher tic severities, ADHD and OCD (Martino & Leckman, 2013).

Communication in Depression and Anxiety. Towbin et al. (2005) found that children with mood and anxiety disorders scored more than one standard deviation below the mean, on average, on the communication composite of the CCC-2 and 32% of children had low social interaction composite scores. Van Steensel et al. (2013) found that children with anxiety disorders have elevated levels of communication challenges compared to nondisabled peers. Halls et al. (2015) found that children with social anxiety disorder were three times more likely to have social communication challenges on the Social Communication Questionnaire (SCQ; Rutter, 2003) than children with other anxiety disorders. There are no studies comparing communication skills in children with TS who do and do not have anxiety disorders; however, it is very possible that an additional diagnosis of anxiety, especially social anxiety, could contribute to communication challenges in children with TS.

In summary, all of the co-occurring conditions discussed above have some association with social communication challenges, and it is reasonable to assume that these conditions may contribute to functional communication challenges in children with TS. In particular, there is substantial evidence that children with ADHD have challenges with not only social communication but also other related language skills (high-level language, discourse, and language formulation). More research is still needed comparing children with pure vs. complex

TS and a focus should be placed on a range of language skills to cover core, high-level, and social domains.

Social Communication in Children with TS

Differences in social cognitive and neuropsychological skills could result in communication differences for children with TS (Hyter, 2007; Wiseman-Hakes 2020). The possible role of social communication in TS has been hypothesized by neuropsychology researchers. Albin (2018) suggested that TS may be characterized as a disorder of the social decision-making neural network (a disorder of social communication). Coming from a neuropsychological perspective, Eddy (2021) reviewed research studying social communication in children and adults, focusing more on adults due to the larger body of current evidence. Eddy drew several conclusions about how pragmatic language (i.e., social communication) may be affected in individuals with TS: (1) Individuals with TS exhibit an over-efficiency with responding automatically to environmental cues, making them quicker with some language tasks (e.g., Walensky et al., 2007). However, this combined with a greater tendency toward difficulties with impulse control, can lead to inappropriate automatic communication responses that go uninhibited (e.g., saying the first thing that pops into your mind). (2) Difficulties comprehending non-literal remarks are present only when the meaning and intention of the speaker differ, but are not present when meaning and intention do not differ (e.g., sarcasm and metaphor; Langdon & Coltheart, 2004). While these challenges may be due to problems understanding more complex mental states, another explanation is that individuals experience ambiguity about the communicative intent of others due to their own experiences of difficulty following social norms when communicating. (3) Individuals with TS may process nonverbal communication information differently on a neural level and this may involve differences in the mirror neuron

system. Eddy weighed the evidence for TS as a disorder that is inherently social (as suggested by Albin, 2018), agreeing that many tics can be seen as social-communicative. Eddy also evaluated evidence for TS as a language disorder of the right hemisphere but concluded that evidence points more toward differences in hemispheric communication, specialization, or dominance. Finally, Eddy considered the idea that TS is associated with differences in social cognition that, in turn, affect pragmatic communication. Eddy concluded that individuals with TS may use more automatic processes, rely on visual information more, and have more complex mental processes to complete when interpreting communication, and suggested that future research on the developmental trajectory of language and social skills in individuals with TS is needed.

Three studies have investigated aspects of social communication in children with TS using parent report questionnaires. Verté et al. (2005) used the original version of the Children's Communication Checklist (CCC, Bishop, 1989) to measure pragmatic language from parent and teacher perspectives (excluding the speech and syntax subscales) in 24 children with TS. Parent and teacher reports alike indicated greater communication challenges for children with TS compared to nondisabled peers. The number of TS participants who also presented with ADHD was not reported; however, the TS group presented with significantly greater inattentive and hyperactive symptoms as a whole compared to nondisabled peers. Since inattentive and hyperactive symptoms were not analyzed with respect to communication scores, conclusions about the contribution of inattention/hyperactivity as a factor could not be reached.

Güler et al. (2015) reported higher scores (indicative of greater challenges) on the SRS for children with TS compared to nondisabled peers. This was observed for the SRS Total Score, as well as the Social Communication subscale, and all other subscales. Fifty-eight percent of the TS sample had a co-occurring diagnosis of ADHD. For the total SRS score, a further analysis

was conducted showing that all of the variance between the TS group and nondisabled peers could be attributed to co-occurring conditions. This covariate analysis was not completed for the communication subscale. Thus, co-occurring conditions appear to have an important contribution concerning social responsiveness in children with TS, but their contribution was not analyzed for communication scores. A limitation here was the small sample size. There were 31 participants, and only five had a diagnosis of pure TS.

Also using the SRS, Darrow et al. (2017) examined parent -social communication in subgroups of children and adults with TS and compared these scores to unaffected family members. Participants who had TS and ADHD had significantly elevated levels of communication challenges. Participants who had pure TS did not differ significantly from the unaffected group. A limitation of the Darrow et al. (2017) study was the use of family members as a control group. This may have resulted in a skewed control sample since TS and other related conditions (e.g., OCD and Autism) run in families. Darrow et al. (2017) suggested that further research is needed to understand social communication symptoms in children with TS and that social communication should be investigated even in individuals with TS who do not meet Autism criteria.

On the whole, the findings of these studies suggest that children with TS have social communication challenges; however, the social communication skills of children with pure TS may not be affected. More research is needed on this topic since only one study to date has considered co-occurring conditions in the analysis (Darrow et al., 2017) and interpretation is limited by the fact that the control group may not have been composed of nondisabled peers. The use of the SRS Social Communication subscale is another limitation of some studies (Darrow et al., 2017; Güler et al., 2015), as it is geared specifically toward identifying the social

communication features that characterize Autism. Future research is needed that collects information about a broader range of social communication skills, as features that characterize communication in Autism may not be the same as those that are important for understanding TS. Future research should also use an adequate sample size, analyze children's data separately from that of adults, compare to samples drawn from the general population, and consider co-occurring conditions in the analysis. Study 2 in this dissertation will address these needs using a series of parent questionnaires.

Social/Psychosocial Functioning in Children with TS

In addition to literature focused on skills underlying communication, the way in which tics are perceived by communication partners may contribute to the social-communicative experiences of individuals with TS. Tics are repetitive behaviors not intended to be communicative; however, because tics largely involve vocalizations and movements of the face, neck, and shoulders, they may be interpreted as communicative by others in the child's environment. Tics may appear more socially relevant when they are complex and, when pragmatically inappropriate, can have deleterious effects on social functioning and social relationships. Some examples of socially inappropriate tics are coprolalia or "swearing tics" and non-obscene socially inappropriate symptoms (NOSIS) (e.g., laughing at inappropriate times and making inappropriate comments about people's appearance). Eddy & Cavanna (2013) estimate that two-thirds of individuals with TS experience NOSIS.

Research has found that children with TS often experience problems with peer relationships and social functioning (e.g., O'Hare et al., 2015). Some factors related to social functioning in school are the child's personality, the severity of their condition, and the behaviours/attitudes of other students. Stigma and bullying associated with TS are common

experiences for children (e.g., O'Hare et al., 2015; Malli et al., 2016). Smith et al. (2015) found that children with TS often have negative experiences with teachers in school. Children with TS may avoid some social opportunities and therefore may have less experience with social interactions compared to other same-aged children. Sukhodolsky et al. (2003) documented general social difficulties in both pure TS and TS+ADHD groups, using the Social Competence subdomain of the Child Behaviour Checklist (Achenbach & Rescorla, 2001). It is important to note that many factors linked to the social experiences of children with TS relate not just to the child's qualities and skills, but to their environment (e.g., the behaviours and attitudes of others in school) and their attitudes about themselves. These factors make up the child's context and must be considered in addition to "within child" factors.

More broadly, psychosocial functioning (which considers social and psychiatric function) is also known to be lower in young people with TS compared to typical peers (Gorman et al., 2010). No research has been conducted investigating the relationship between psychosocial functioning and communication skills in children with TS. The present research will address the relationship between these two constructs, hypothesizing that they are positively correlated.

Summary and Areas for Future Research

The information provided above demonstrates how social cognitive and neuropsychological factors, language development, co-occurring conditions, and social behaviours/experiences all contribute to the communication experiences of children with TS. These various, interrelated factors produce a complex picture of communication for children with TS. Interest in social communication in TS has been a recent development in the field of TS research (i.e., Albin, 2018; Eddy, 2021).

Thus far, there have been no studies of the day-to-day communication functioning of children with TS. The specific nature of communication challenges that children with TS experience is under-researched. The complex issue of the influence of co-occurring conditions is an important consideration in this research as several of these conditions are known to come along with their own communication challenges. The difficulties of children with ADHD are the most well-studied currently. To date, there is some evidence of greater communication challenges in children with co-occurring ADHD; however, most studies have not systematically addressed this issue by collecting information about other co-occurring diagnoses and considering them in relation to communication skills. Given that children with complex TS are at a much higher risk for communication concerns, detailed information about their communicative function is of particular interest to speech-language pathologists (SLPs).

Dissertation Purpose

The overarching aims of this research are to 1) determine what research exists currently on communication skills in children with TS, 2) contribute to the literature about the proportion of children with TS who experience communication challenges and whether they have co-occurring conditions, 3) understand what communication domains present challenges for children with TS, and 4) understand real-world contextual factors that relate to functional communication among children with TS. These aims are addressed through three studies. Study 1 is a scoping review, Study 2 is a parent-reported measures study, and Study 3 is a mixed methods case series with five families.

Study 1

Study 1 is a scoping review of the literature that aimed to describe what is known about the language and social communication skills in children with TS and to identify the gaps in the literature.

Study 2

Study 2 is a parent-reported measures study that investigated rates of communication challenges and examined individual domains of communication in 61 children with TS by looking at parent reports across a subset of children with a variety of profiles of TS. It addressed the relationship between communication skills and overall level of function, using a measure of psychosocial function. Study 2 also considered the severity of the tic disorder in relation to communication functioning and overall functioning. I hypothesized that scores for communication domains involving social communication, coherence in communication, and high-level language would be lower compared to a same-aged normative sample. Study 2 also addressed children's co-occurring diagnoses in relation to measured communication skills. Although past findings have been conflicting, ADHD is theoretically expected to be related to lower communication skills and thus, I hypothesized that children with TS with a diagnosis of ADHD would have lower scores compared to children without co-occurring ADHD. Finally, in this study, I also sought to compare the communication skills of children with and without anxiety and OCD, recognizing that this analysis was exploratory due to the limited current evidence.

Study 3

Study 3 is a mixed methods case series focused on gaining an in-depth understanding of communication development in five children who have experienced challenges with their

language development. It aimed to understand communication development using a detailed battery of testing (quantitative data) mixed with data focused on the day-to-day communication functioning of children (qualitative data). Contextual factors related to communication success were considered by collecting information about communication from parents' perspectives. I hypothesized that children would run into specific challenges in their day-to-day communication interactions and that the environment/context would play a role in facilitating or hindering their success in specific interactions.

Research Questions

To address the study aims, there are seven research questions:

Study 1

1. What is the scope and landscape of the current literature on language and social communication skills in children with TS?
2. What can we conclude about language and social communication skills in children with TS?
3. What research is needed to understand language and social communication in children with TS?

Study 2

4. Do parent-reported communication skills in children with TS differ from a normative sample?
5. How do co-occurring conditions predict general communication and social-pragmatic communication scores in children with TS? In particular, to what extent does an additional diagnosis of ADHD predict communication skills in children with TS?

6. What is the relationship between communication functioning and levels of parent-reported psychosocial functioning in children with TS?

Study 3

7. What is the nature of communication challenges in children with TS who have experienced challenges in their communication development and how is communication facilitated or impeded in day-to-day functional communication contexts?

Theoretical, Philosophical, and Methodological Frameworks for Mixed Methods Research

Mixed methods research provides a method of combining two very different viewpoints. In this thesis, Chapter 5 combines Anglo-American and the Continental perspectives on communication through quantitative methods (to gather information from an Anglo-American perspective) and qualitative methods (to gather information from a Continental perspective). This section will describe the Bioecological Model, which allows for the integration of aspects of the child's communication system (Anglo-American view on commutation) and aspects of the child's day-to-day contexts (Continental view of communication). Functionalism, the theory used to frame the Continental view on communication, is also described. Next, this section will demonstrate how methodological coherence has been on interpretive description and naturalistic inquiry. The background provided here includes a discussion of the scaffolded methods: case study research, interpretive description and naturalistic inquiry. Finally, this section will discuss interpretive frameworks (the overarching theoretical standpoint) and researcher positionality/reflexivity.

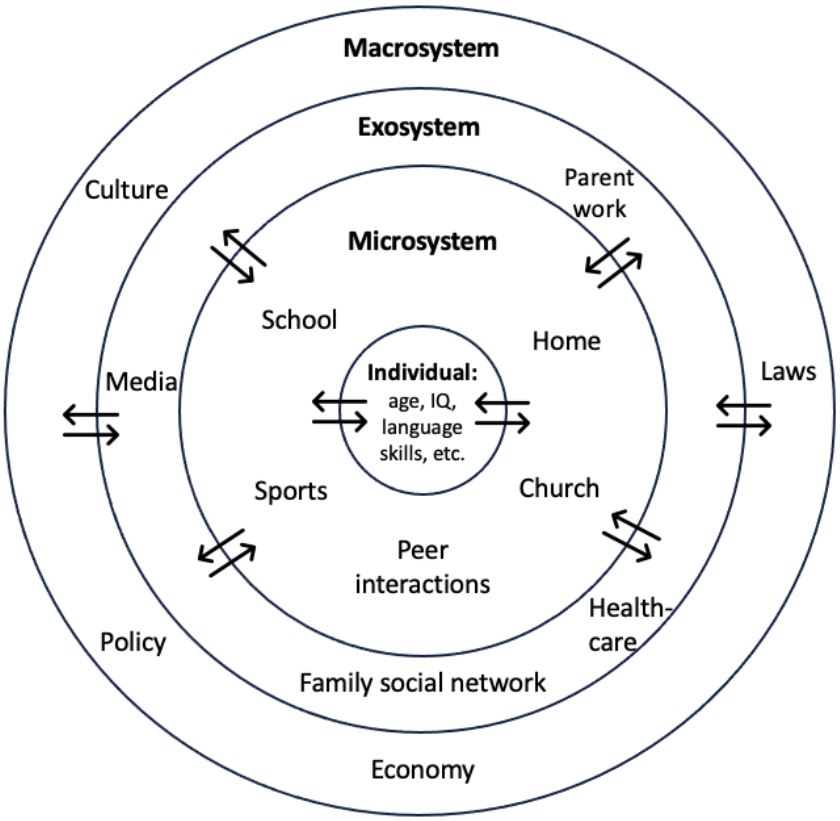
The Bioecological Model

Urie Bronfenbrenner's bioecological model (also termed social-ecological model/ ecological systems theory) is a theoretical model used to explain childhood development

(Bronfenbrenner, 1979). It helps to articulate how different levels of contextual factors may relate to communication. The bioecological model sees development as a product of interactions between an individual and their many world contexts, such as day-to-day interactions with others, family, geographical location, cultural norms, healthcare, and political landscape. At the level of the individual, cognitive, biological, and socio-emotional factors interact with one another. Additionally, all of the world-context factors interact with one another, creating a complex system of effects. This model distinguishes between different levels of context using terminology: micro-, exo- and macro-system (see Figure 1). The bioecological model contributes to the current research by highlighting that the course of individual development (in this case communication development) is shaped by complex interacting factors on many contextual levels. This research placed a focus on the "individual" and "microsystem" by using direct observation of the child and information from the parent.

Figure 1

Adaptation of the Bioecological Model Figure (based on Zhou & Brown, 2015)



Functionalism

Functionalism is a perspective that sees language use/function as an overarching factor, sitting above other levels of the linguistic system (i.e., language structure; Bates, 1991). Functionalism was championed by Carol Prutting (1982; Prutting & Kirchner; 1987; Bates, 1991) When considering language development, the functional perspective emphasizes that communicative development is governed by the expression and comprehension of communicative intent. This is in contrast to a structuralist perspective which emphasizes the separation of structure and function (as in the Bloom and Lahey form, content, and use model; 1978). Functionalism is common in the linguistic study of social communication and suggests that contexts must be preserved when studying language (Hyter, 2007; Barnes & Bloch, 2019; Matthews, 2014; O'Neil, 2012; Perkins, 2014).

Functionalist language research also supported the social constructionist view of language development pioneered by Lev Vygotsky. I have selected the functional perspective because emphasizing function facilitates the preservation of context in the study of language, which is particularly important for studying communication in children with TS due to the potential effects of environment/context on communication. This perspective facilitates uncovering real-world factors connected to successful and unsuccessful day-to-day communicative interactions. The functionalist perspective was applied by focusing on the child's real-world comprehension and expression of communicative intent. This was done by asking the parent to record specific and detailed examples of successful and unsuccessful communication in a log book and asking the parent about these examples and other observations/perspectives of functional communication in the parent interview. The functionalist perspective and the bioecological model complement one another in this research. While the functionalist perspective places importance on real-world communication successes and challenges, the bioecological model recognizes that contexts affecting communication are interdependent and dynamic.

Case Study Research, Interpretive Description, and Naturalistic Inquiry

Mixed methods case studies combine quantitative and qualitative data using both induction and deduction to understand complex topics (Creswell & Plano Clark, 2018; Harrison, 2017). Case study research is beneficial when there are many variables at play and was ideal for gaining an in-depth understanding of communication for a small number of children with TS who have many complex social, psychological, and biological factors at play. Each case is defined by the boundaries that are placed around it (e.g., it is studied within certain contexts or focusing on a certain topic). Chapter 5 draws on the case study approach of Robert Stake (1995)

by using a constructionist worldview (described below) and focusing on the unique features of particular cases that the researcher can learn from.

Interpretive description is a qualitative approach developed for applied research (Thompson Burdine et al., 2021; Thorne, 2016). Interpretive Description aims to gain a deeper view of the perspectives of participants (in this case how parents see their child's communication and the contextual factors at play). It focuses on answering clinical questions within health disciplines. Interpretive description does not aim for "saturation" like some qualitative paradigms because Interpretive Description highlights that it would be difficult or impossible to understand all perspectives and experiences because they are so numerous and varied. Interpretive description often involves qualitative interviews but also supports the use of "novel data," or any additional data that can encourage an understanding of the topic (Thompson Burdine et al., 2021, p. 340). Interpretive description encourages an inductive, bottom-up approach to coding data; therefore, codes are not decided a priori. The researcher brings an active analytic lens while developing themes and interprets the data from their own (clinical) perspective while focusing on information relevant to the research question.

Naturalistic inquiry attempts to understand a phenomenon in the settings/contexts in which it naturally occurs (Lincoln & Guba, 1985). Naturalistic inquiry encourages carefully chosen samples, the use of qualitative methods, inductive approaches to data analysis, and findings to be interpreted by the researcher with the use of 'tacit' knowledge (or intuition) rather than being taken at face value using measurement instruments. This is because no instrument is capable of understanding and drawing conclusions about nuanced and complex phenomena that can be interpreted in multiple ways. In reporting, a focus is placed on the particularities of cases rather than on producing sweeping generalizations.

Interpretive Frameworks

An interpretive framework (as defined by Creswell & Poth, 2018) is the overarching or organizing framework that packages the philosophical (ontological and epistemological) assumptions of the research. Two interpretive frameworks provided the lens for these studies, pragmatism and social constructionism.

Pragmatism: An Interpretive Framework for Mixed Methods Research

Pragmatists believe that both quantitative and qualitative research methods can be combined to answer applied research questions and aim to select the methods that best fit the research questions, regardless of differing epistemologies. That is, methods should be selected based on "what works" (Creswell & Poth, 2018). Pragmatism uses a utilitarian perspective to address arguments brought forward by theoretical purists who state that ontological and epistemological perspectives cannot change within a single study. According to pragmatists, combining objective and subjective knowledge to answer research questions is practical (Creswell & Plano Clarke, 2018). In this project, pragmatism provides a rationale for using both quantitative and qualitative methods in a single study, where quantitative methods provide background information about participants' language development from a traditional speech-language pathology viewpoint and qualitative research methods provide a rich investigation of specific communication examples and contextual factors at play.

Social Constructionism: An Interpretive Framework of Qualitative Research

The social constructionist framework views knowledge as a fabrication, built through social systems such as language, culture, and socio-political context. Truth and reality are seen as multiple, subjective, and changing rather than singular, objective, and static (Crotty, 1998). This view is the foundation of Lincoln and Guba's qualitative data analysis method, naturalistic

inquiry (Lincoln & Guba, 1985), whereby the researcher collects information about a phenomenon of interest in its naturally occurring setting. Social constructionism is the interpretive framework guiding the qualitative interpretive description in this research.

Researcher Positionality and Reflexivity

I have used my clinical background in the field of speech-language pathology, my personal experiences with neurodivergence, my perspective as a member of a mental health team in a TS clinic, and my perspectives as a family member of a person with complex TS as interpretive lenses for this work. Due to its clinical application, interpretive description emphasizes that reflexivity must extend beyond researchers themselves, considering the biases inherent in the clinical discipline as well (Thorne, 2016).

I have reflected on speech-language pathology as a discipline and how my clinical background shapes the questions I ask as a researcher and molds my thinking about what can be learned about communication in children. Speech-language pathology is a discipline built on the medical model, which is primarily a deficit-based model. There is an assumption within the discipline that being able to talk clearly, form sentences correctly, get your point across using spoken language, and understand the spoken language of others is beneficial and desirable for individuals in their day-to-day lives. Pediatric speech-language pathology is interested in language development as a process and assumes that if an aspect of communication development is lagging behind age-based expectations, something can be done (and should be done) to fix it. I have attempted in this research to move away from deficit-focused thinking and toward not problematizing differences. This shift naturally leads to a perspective that is open to accepting differences, rather than changing or "fixing" them.

On both personal and professional levels, I have had a variety of experiences with working with, interacting with, and caring for neurodivergent people. I recognize neurodivergent people, like those with TS, as people who will look and/or act differently from what is considered "normal" no matter what interventions they receive or how hard they try to be different from how they are. Neurodivergent people are faced with a unique challenge that is not easily understood by helping professionals who lack personal experience with neurodivergence: finding a way to function in a society that sees you as flawed and navigating a delicate balance between working harder than everyone else and conserving your energy, between acting to keep others feeling comfortable and being your authentic self, and between conforming and radically accepting yourself as you are. Coming from this perspective, I hope that any differences in language skills that I find are treated with care. When we find differences, I believe that we must always ask, "Does this matter, and why?" ("Is there a *functional* reason why we should focus on this difference?"). I will report the differences that I observe in children with TS in this research but I intend to point out both strengths and weaknesses. I also intend for the information that I share to start a conversation about whether or not the differences matter for day-to-day functioning, and if they do, what can be done to provide support. This could mean coaching and skill building, as we traditionally offer in speech-language pathology. But it could also mean educating others to be more accepting of differences, changing demands on individuals who are different, and universal designs with specific differences in mind.

Because of my work with children in the TS clinic, I came to this research with an assumption that the communication challenges of children with TS are frequently overlooked by parents and healthcare professionals due to complex medical and neuropsychological presentations and the fact that challenges can be subtle or specific to only one area. Given these

past experiences, I needed to stay aware of my assumptions and be open to the idea that parents may have different levels of understanding about their child's communication challenges, that I might see a wide range of language skills in the children, and that different methods of data collection may or may not tell the same story about each child's skills.

Being a family member of a person with TS and interacting with parents who have children with TS has given me a more holistic view of communication functioning in these children. These children are extremely diverse and their families have complex lives. An important aspect of my reflexivity was to consider all of the aspects of the cases that I encountered, not just the experiences that parallel my own as a family member of someone with TS.

Chapter 2: A Scoping Review of Language and Social Communication Abilities in Children with Tourette Syndrome

This study has been published as: Feehan, A. & Charest, M. (2024). A Scoping Review of Oral Language and Social Communication Abilities in Children with Tourette Syndrome. *International Journal of Language and Communication Disorders, 59*(1), 143-164.

Since the time of publication, our use and understanding of anti-ableist language has evolved; therefore, for the purpose of this document, the following changes have been made:

- 'oral language' changed to 'language,'
- 'control group/controls' changed to 'nondisabled peers,' and
- 'typically developing' changed to 'nondisabled.'

The original terms remain in the published journal manuscript.

Abstract

Background. Children with Tourette syndrome (TS) have historically experienced problems in academic and social settings, yet their language and communication abilities have not been extensively researched.

Aims. This scoping review maps the literature on the language and social communication abilities of children with TS in order to describe the nature of the current literature, present a summary of major findings and identify where gaps exist.

Methods. A scoping review was completed to identify studies measuring the language or social communication abilities of children with TS. A systematic search of six electronic databases was conducted to obtain published and unpublished literature. All English studies measuring the language or social communication abilities of children with TS were included. Information was extracted from records and knowledge was synthesised in a narrative summary.

Main Contribution. We identified 56 records for inclusion. Almost all records were located in journals within the fields of psychology and psychiatry. Skills most often studied were verbal IQ and verbal fluency. The literature suggests an increased prevalence of language disorders and social communication problems in children with TS; however, literature comprehensively detailing these challenges was scarce. Language strengths were identified in verbal intelligence, story/sentence recall, categorisation and performance on tasks at the single-word level.

Conclusions. Language and social communication skills are important for academic and social success. This review brings scattered literature together to provide up-to-date information about language in children with TS and highlights that there are considerable gaps in our knowledge about language and communication in this population. This scoping review can

inform future research and support speech-language pathologists in the assessment of young people with TS.

KEYWORDS language development, language disorders, scoping review, social communication, Tourette syndrome

WHAT THIS PAPER ADDS

What is already known on the subject: Speech-language pathologists (SLPs) working in various contexts (e.g., schools, mental health teams) are likely to encounter children with Tourette syndrome (TS); however, the description of this population and potential communication characteristics is not well represented in the speech-language pathology literature. Previous literature reviews have reported strengths in verbal fluency and morphological processing. Challenges in expressive language, higher order language, social cognition and a propensity towards autistic traits have also been identified.

What this paper adds to existing knowledge: This review differs from previous narrative reviews by employing a systematic approach to searching for literature. As a result, we identified 25 additional studies that had not been cited in previous reviews and additional relevant findings in 23 previously reviewed studies. This review confirms several previous conclusions about language in children with TS and extends or clarifies several others, thereby providing the most current information on language and social communication abilities. The use of current taxonomies of language and social communication helps to organise this literature for clinicians and researchers in speech-language pathology and identifies a need for further research from the speech-language pathology perspective.

What are the potential or actual clinical implications of this work? · These results imply that SLPs should screen children with TS for language disorders and investigate social communication and social interaction development. Clinicians can expect greater challenges in language and communication development for children with complex forms of TS (i.e., those who exhibit co-occurring conditions such as attention-deficit/hyperactivity disorder). The multidisciplinary nature of the current literature implies that clinical collaboration with other disciplines will be of particular benefit to serving this group of children.

Introduction

Tourette syndrome (TS) is a neurodevelopmental disorder involving one or more vocal tics and multiple motor tics (American Psychiatric Association, APA, 2013). Tics can be simple, such as grunting (vocal) or eye blinking (motor), or complex, such as saying words (vocal) or completing a series of gestures (motor). TS is associated with several neurobiological and neurochemical differences, including reduced caudate volumes, cortical thickening, increased volumes of the corpus callosum (Greene et al., 2013) and dysfunction of the dopaminergic system (Singer, 2013). The prevalence of TS in children is estimated to be between 0.2% and 0.3% (Bisko et al., 2022), and boys are affected more than girls by a factor of 3.5 (Baizabal-Carvallo & Jankovic, 2022). Tics begin in childhood, peak in severity between 10 and 12 years of age and wane in adulthood (Bloch, 2013). In about 80% of cases, TS co-occurs with one or more co-occurring conditions, such as attention-deficit/hyperactivity disorder (ADHD), obsessive-compulsive disorder (OCD) and anxiety and depression (Martino & Leckman, 2013). Quality of life has sometimes been reported as low for individuals with TS (e.g., Marek, 2006).

Motivation and Background to the Review

Existing literature reports difficulties in areas of academic, neuropsychological and social function in children with TS and subtle challenges in aspects of social cognition in adults with TS. These areas of noted difficulty are all closely connected to language and communication function and therefore invite a closer look at language and communication skills directly. A summary of findings in these areas of development provides context for the review.

Summary of Academic, Neuropsychological, and Social Development in Children with TS

Children with TS have been found to have high rates of learning disabilities and these challenges sometimes are in areas of language development (i.e., writing, spelling and reading;

Claussen et al., 2018; Mahone et al., 2002). Despite this, the relationship between language learning and learning disabilities in children with TS has received surprisingly little research attention.

Investigations into the neuropsychological characteristics of children with TS have explored aspects of cognition that are important for organising and regulating communication interactions, specifically, attention, memory and executive functioning (Hyter, 2017). Findings in children with TS point to challenges in divided and sustained attention (Chang et al., 2007; Sherman et al., 1998), long-term nonverbal memory (e.g., Bloch et al, 2006), inhibitory control (Morand-Beaulieu et al., 2017a) and executive control (Sikora et al., 2019).

Several studies have found that children with TS experience lower social functioning compared to peers without TS (Stokes et al., 1991; Sukhodolsky et al., 2003; Zhu et al., 2006). When rated by their peers, children with TS may be perceived as more withdrawn, more aggressive and less likable (Bawden et al., 1998; Stokes et al., 1991). Some studies have reported difficulties in making and keeping friends and developing secure attachments (Champion et al.,1988; O'Hare et al., 2015). Many factors are likely to influence the social experience of children with TS; however, the presence of problems in social function indicates that we need to know more about how children with TS manage communication in social interactions. Social communication skills are necessary in social interactions and therefore tie in closely with social functioning.

Social Cognition in Adults with TS

Individuals draw on social cognitive skills during social communication and these skills are often considered foundational to social communication (e.g., Adams., 2005; Hyter, 2017). The research programme led by Eddy and colleagues in the early 2010s (e.g., Eddy et al., 2010a,

2010b, 2011) has documented that adults with TS process social information differently (see Eddy et al., 2017). Furthermore, socially inappropriate behaviour is a well-known aspect of TS (i.e., cursing tics and non-obscene socially inappropriate symptoms; NOSIS), which, along with social cognitive challenges, may indicate that pragmatic language challenges are a core feature of TS (Albin, 2018; Eddy, 2021; Eddy & Cavanna, 2013; Eddy et al., 2011). Studies of adults with TS have reported subtle challenges in several aspects of social cognition that tie into social communication, including understanding the appropriate use of nonliteral language (i.e., sarcasm and metaphor), reading intentions, understanding humour, understanding emotional states, understanding and considering others' perspectives or mental states, social problem solving, social reasoning and recognising social indiscretions (Channon et al., 2003a; Channon et al., 2012; Eddy & Cavanna, 2015; Eddy et al., 2010a, 2010b, 2011, 2015). Across these studies, co-occurring diagnoses were present in a minority of participants, indicating that social cognitive differences may be central to TS. There are also some studies reporting no differences compared to nondisabled adults on social cognitive tasks involving theory of mind, empathy, identification of mental states and comprehension of emotional cues (Baron-Cohen et al., 1997; Channon et al., 2004; Devinsky et al., 1993). The nuances of specific task demands and task difficulty may help to explain these inconsistent findings. Nonetheless, the accumulated findings suggest that social cognitive skills need to be explored more thoroughly in children with TS as they are relevant to social communication development in childhood.

Previous Reviews of Language and Social Communication in TS

These observations of challenges in aspects of social and cognitive functioning suggest that speech-language pathologists (SLPs) may have a role in supporting language and communication in individuals with TS. This has motivated us to gather and organise literature

that considers the full scope of language and social communication from a speech-language pathology perspective. Knowledge on this topic can serve to highlight the potential role of SLPs in supporting children with TS and can help direct future research.

Several previous reviews have addressed aspects of language and communication in TS (Burd et al., 2008; De Nil et al., 2006; Morand-Beaulieu et al., 2017b, Murphy & Eddy, 2013), with the most recent being a book chapter focused on social communication (Eddy, 2021). Eddy (2021) summarised several studies of language and social cognitive abilities, and outlined how tics and other behaviours in TS can violate conversational rules and interfere with communication. Eddy highlighted the need for a greater understanding of social challenges in TS from a pragmatic point of view, discussing several theories on the relationship between TS and social communication.

Earlier reviews have suggested that expressive language may be an area of challenge for individuals with TS (Burd et al., 2008; De Nil et al., 2006; Murphy & Eddy, 2013); however, specific strengths in expressive language have also been reported: verbal fluency, stem completion, naming and past tense production speed (Morand-Beaulieu et al., 2017b; Murphy & Eddy, 2013). Language comprehension has also been suggested as a challenging area (Burd et al., 2008; Morand-Beaulieu et al., 2017b), with a specific need identified for studies examining complex language comprehension (Murphy & Eddy, 2013). Difficulties on measures of high-level and non-literal language have been identified (De Nil et al., 2006; Morand-Beaulieu et al., 2017b; Murphy & Eddy, 2013). Finally, past reviews have noted several factors that may relate to language and communication skills. These include motor skills, medication use, tic severity, social cognitive problems, inhibition and retrieval problems, the presence of autistic traits, co-

occurring ADHD and other co-occurring conditions (Burd et al., 2008; De Nil et al., 2006; Morand-Beaulieu et al., 2017b; Murphy & Eddy, 2013).

These prior reviews provide important insights into language and communication in TS; however, none reported a systematic search strategy and none provided the broad focus that characterises speech-language pathology models of language and communication. Moreover, while knowledge specific to childhood is relevant for developmentally-focussed clinicians and researchers in speech-language pathology, prior reviews have not focussed explicitly on children. The current scoping review differs from previous reviews in its focus on studies of children only, its intentional search for a wide range of measures of language and social communication, its use of a systematic search to capture relevant studies, and its focus on organising findings from a speech-language pathology perspective. Some of the results of the current review overlap with findings from past reviews: Table 1 provides a summary of this overlap. To preview the results, the present search identified an additional 25 papers that were not described in previous reviews. Additionally, finding details that were not reported in previous reviews have been added in this scoping review.

Table 1

Previous Reviews of Language and Communication in TS: Coverage and Gaps

	Number of studies cited (Language findings partially reported/ not reported)	Number of studies cited (Complete language findings reported)	Number of studies *not* cited
Eddy (2021)	8	4	44

	Number of studies cited (Language findings partially reported/ not reported)	Number of studies cited (Complete language findings reported)	Number of studies *not* cited
Morand-Beaulieu et al. (2017)	18	7	31
Murphy & Eddy (2013)	2	3	51
Burd et al. (2008)	1	0	55
De Nil et al. (2006)	9	1	46
***Total**	**23**	**8**	**25**

*Excludes overlap across reviews.

Purpose and Objectives of the Current Scoping Review

The purpose of the present review was to locate and describe the literature on language and social communication in children with TS. For our purpose, we define language as oral skills of expressive/receptive language, semantics, morphosyntax, narrative language and high-level language. In keeping with Adams (2005) taxonomy of social communication, we define 'social communication' as skills of social interaction, social cognition, pragmatics (verbal and nonverbal) and language processing. The scoping review does not address characteristics of tics, speech sound production or fluency in TS. For information on these topics, readers are directed to De Nil et al. (2006), Donaher (2006), Krah (2002) and Kwak and Jankovic (2002).

The objectives of this review are (1) to identify the scope and landscape of the current literature on language and social communication abilities in children with TS, (2) to present a

summary of findings, and (3) to identify where gaps exist in this literature. A scoping review was chosen as the methodology as it can facilitate answering broad questions about the existing literature, such as 'what is known about this concept?' (Tricco et al., 2018, p. 467). A scoping review is also suited to mapping a literature that is scattered across several disciplines.

Methods

The protocol for this review was drafted using Preferred Reporting Items for Systematic Reviews and Meta-analysis Extension for Scoping Reviews (PRISMA-ScR, Tricco et al., 2018). We followed the framework introduced by Arksey and O'Malley in their seminal paper (2005) and the advances to this methodology introduced by Levac et al. (2010). We were informed by the systematic process for scoping reviews outlined in Peters et al. (2015).

Literature Search

Our search for literature began with a search strategy developed by the two authors and with the support of library services at the University of Alberta. A three-step process as described by Aromataris and Riitano (2014) and Khalil et al. (2016) was employed, and several iterations were required before the full search could be carried out. First, a list of keywords relating to language and communication was developed based on terminology commonly used by SLPs and language researchers. Search terms were chosen to broadly capture literature about language and social abilities in people with TS. This list was refined through an initial review of literature in CINAHL and MEDLINE. Second, index terms were identified in each electronic database. Third, hand searching of reference lists of all papers that remained after applying our exclusion criteria was completed. See Appendix A for the full list of search terms and an example of our search strategy in MEDLINE.

To locate published documents, the following six databases were searched with adaptations to the search terms made as needed to meet database-specific requirements: OVID MEDLINE (1946-present), OVID EMBASE (1974-present), EBSCOHost CINAHL Plus with fulltext (1937-present), EBSCOHost ERIC, EBSCOHost Education Research Complete, EBSCOHost PsycINFO. In order to identify unpublished literature, two further databases were searched: Bielefeld Academic Search Engine (BASE) and ProQuest Dissertations & Theses. Searches were conducted on 9 February 2022. Results were exported into RefWorks. Duplicates were identified using the RefWorks duplicate tools and through hand searching. Duplicates were reviewed item by item and removed, leaving 865 records.

Inclusion Criteria

We included (1) scientific studies reporting primary data (i.e., information targeted at the general public was excluded; reviews and summaries were excluded, but their reference lists were searched for relevant papers), (2) studies that included children with TS (in some cases groups with co-occurring conditions were present in the study; studies focussed on other tic disorders were not included), (3) Studies that focussed on children 6–18 years of age, and (4) studies that investigated concepts of oral linguistic ability or social communication.

Disagreements throughout the process were resolved by consensus. Only papers written in English were included. No specific date limits were applied and no exclusion criteria regarding methodology or number of participants were applied. Older research was included to be thorough and to allow for a full appraisal of what has been studied in the past.

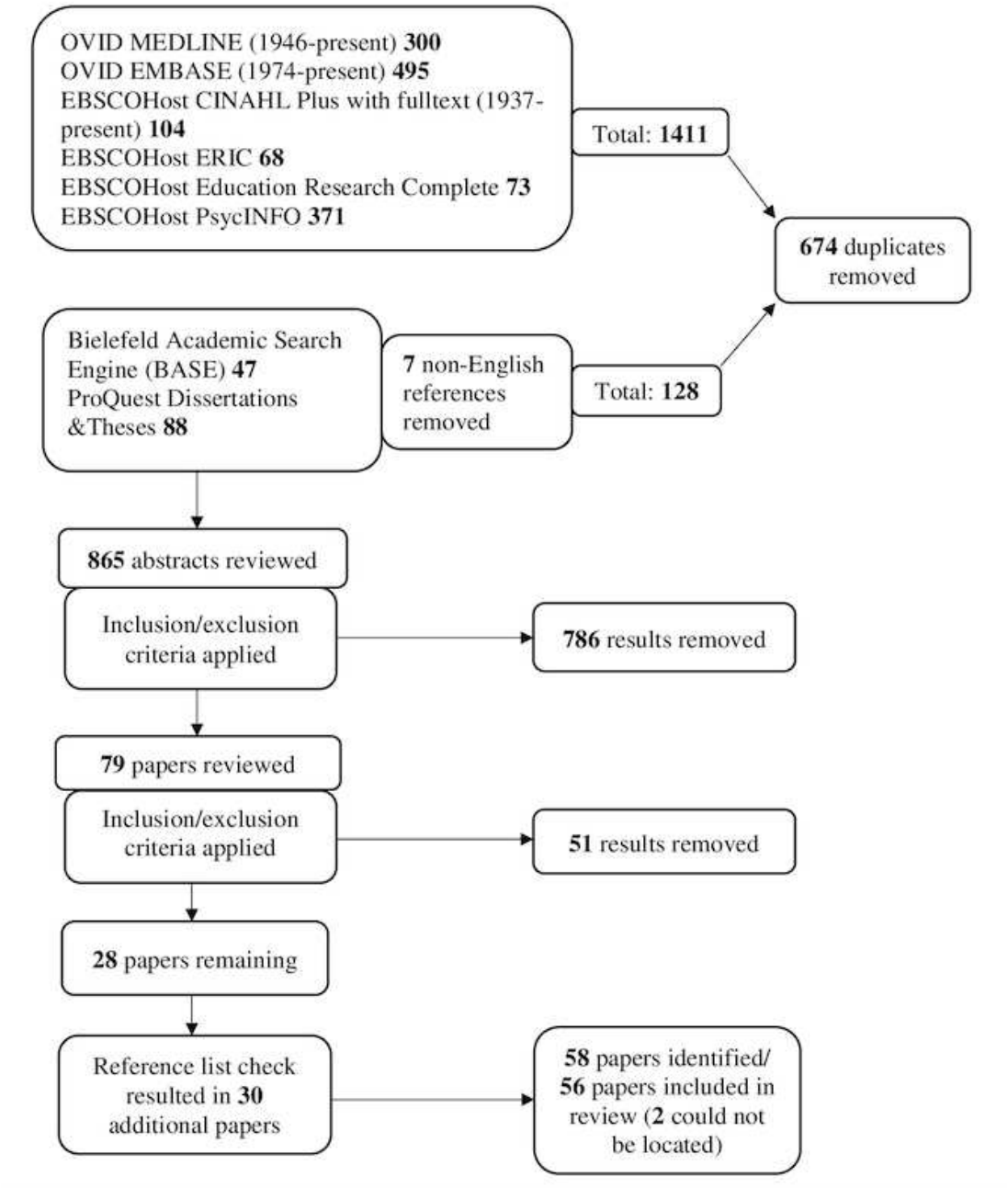
Title/Abstract Screening

The two authors independently reviewed all 865 titles and abstracts. Where there was ambiguity about any of the inclusion criteria, the paper was carried forward for a review of the

full text. Inclusion and exclusion criteria were refined throughout the title and abstract review process. Search terms were broad enough to capture language studies involving the linguistic aspects of tics and studies involving reading and writing, however, these studies were excluded as we refined the boundaries of our topic area (language and social communication). At the title/abstract screening stage, 786 records were excluded, leaving 79 remaining. Figure 2 outlines the flow of search for records.

Figure 2

Flow of Search for Records



Full Text Review

For the remaining 79 articles, a full text review was completed independently by the two authors. Inclusion/exclusion criteria needed to be further defined at this stage. This was done by

defining what constituted a study of children, what constituted a measure of language, and what constituted a measure of social communication.

Studies that involved both children and adults were included only if the population consisted of mostly children (>50% 19 years or younger) or if results for the paediatric population were presented separately.

Studies were included if they reported prevalence of language disorders, case descriptions of language development, receptive and/or expressive language ability, or performance on any task requiring language generation. Neuropsychological assessment tasks (i.e., measures of intelligence, memory and executive function) sometimes involved generating language. All subtests involving categorising, defining/describing and vocabulary knowledge/retrieval were included. Verbal IQ composite scores were also included in the review, as they are derived from language-based tasks. Memory tasks that involved remembering a single word were not included; however, memory tasks requiring participants to reproduce a sentence or story were included, as they are similar to sentence repetition and story retell tasks used by SLPs. From executive functioning studies, we included verbal fluency tasks as they require language retrieval.

We used the Adams (2005) taxonomy to define the parameters of social communication measures. The Adams taxonomy includes (1) social interaction, (2) social cognition, (3) pragmatics (verbal and nonverbal) and (4) language processing (i.e., receptive and expressive language). Therefore, all measures that fell within these four areas of social communication were included (note that language processing measures had already been included under the umbrella of 'language'). All measures that provided an overall social communication score were included as well. Measures were included if they primarily investigated social interaction, defined by us as

the child's ability to interact in social situations or motivation for social interaction. Under social interaction, we also included papers looking at 'social behaviour', 'social incompetence', 'social motivation', 'socialisation', and 'social skills' if the focus was commensurate with our definition of social interaction. Social skills and other social measures were not included if the focus was outside of this definition (e.g., involvement in social activities, quantity and quality of social relationships). We included measures that tested the child's ability to understand facial expressions and prosody but did not include simple facial recognition tasks. Studies measuring social cognitive abilities such as understanding mental states or theory of mind were also included. Finally, studies focussed on autism trait investigation were included, as these measures have a primary focus on social communication/interaction.

Twenty-eight studies remained after this step. Through reviewing the reference lists of these 28 studies and 17 related articles, 30 additional papers were identified for inclusion, for a total of 58. Two of these papers could not be located, leaving 56 records in the review.

Coding and Summarising Articles

Relevant information was extracted from the 56 studies using a spreadsheet developed and refined in collaboration by the authors. Data were fully charted by one reviewer with the second reviewer independently charting 20% of the data. Consistency was 96%. Most disagreements related to multi-part studies where a control group was used for some studies and not others. One author coded them as 'no control group' and the other coded them as 'control group'. All disagreements were resolved by consensus and remaining data were checked for similar differences. The data items that were charted included article characteristics (publication year, author, title and source), study design, population descriptors (total N, age, sex, diagnosis of study groups), language or social communication skill measured and results of language and

social communication measures. There was considerable variability in the measurement tools used across studies and many of them come from disciplines outside of speech-language pathology; therefore, specific tools were not charted. A focus on the skill measured (rather than the specific tool) facilitated the grouping of studies based on language and social communication constructs. In order to synthesise findings, a narrative summary was prepared by (1) grouping studies based on the skill measured; (2) further subdividing studies based on findings and the groups included; and (3) composing a summary of findings for each skill area by reporting on studies that did not consider co-occurring conditions in their groupings, followed by reporting on studies that did consider co-occurring conditions in their groupings.

Results

Time Frame and Methods

Publication dates ranged from 1974 to 2019. Eleven studies were published before 1990, 16 between 1990 and 1999 and 29 after 2000. Studies were categorised as either descriptive (21 studies; 38%) or observational (35 studies; 63%). Descriptive studies included case studies of individuals with TS, studies comparing performance of children with TS relative to norms (normative studies), survey studies and mixed methods with qualitative interviews. Observational studies included comparisons of recruited samples of children with TS and control groups of children (correlational studies).

Sources of Records

The majority of the included records (47; 84%) were published in academic journals (date range = 1974–2019). Six (11%) of the studies were published in dissertations (date range = 1986–2006) and three (5%) were published in books (date range = 1982–1988). Thirty-five different journals were listed for the 47 journal publications, leaving little overlap in the location

of these articles. The most frequent journals were the Journal of Child Psychology and Psychiatry and Allied Disciplines (three articles; all published in 1988) and the Journal of the American Academy of Child and Adolescent Psychiatry (three articles; date range = 2000–2017). Thirty-nine (83%) of the 47 articles were published in journals focussing on psychology/psychiatry, with an additional five (11%) focussing on paediatrics/development, one (2%) focussing on clinical linguistics, one (2%) focussing on autism and one (2%) focussing on genetics.

Sample Characteristics

The size of the TS sample recruited was >30 for 28 studies (50%) and <30 for 27 (48%). One study included multiple samples; one with more than 30 children and one with fewer than 30 children (2%). Participants across studies varied widely in age, from 6 to 67 years of age. Seven studies (12%) included individuals with TS over the age of 19 (in addition to the child participants; recall that our inclusion criteria required that >50% of participants be 19 years of age or younger). Sex breakdown of participants with TS most often included a greater number of males than females (in 53 studies or 95%; including seven studies with only male participants). One study (2%) included a greater number of females than males and two (4%) did not provide a sex breakdown.

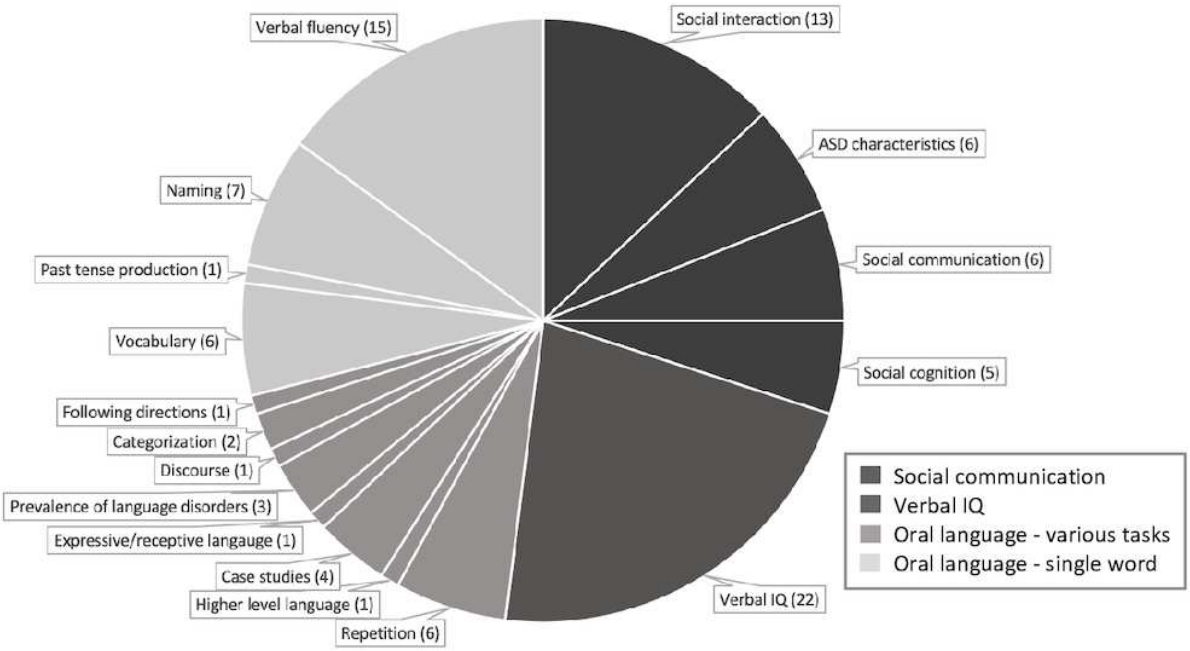
Synthesis of Study Findings

Figure 3 presents a broad categorisation of language and social communication skills measured across studies. For a list of all studies included in the review and a detailed breakdown of the skill measured, see Appendix B. In the narrative summary that follows, where studies described performance in relation to co-occurring diagnoses, we include those findings. We have

used a star (*) throughout our summary to indicate where individual findings have been previously presented in a published review of language in TS.

Figure 3

Language and Social Communication Skills Measures Across Studies



Language Findings

Twenty-three studies reported on the language abilities of children with TS. Of these, 16 studies had not previously been discussed in a review of language in TS. The prevalence of language disorders in children with TS was reported in three studies. Claussen et al. (2018) reported the prevalence of parent-reported speech/language conditions as 30% in a US national sample of children with TS, compared to 4% in a sample from the general population. In the TS sample, 80% of children had a co-occurring condition; 55% had co-occurring ADHD. The occurrence of speech/language conditions was not reported in relation to these co-occurring

conditions. Cravedi et al. (2018) examined the prevalence of language delays/disorders in different clusters of children with TS. Prevalence was 45% in children with 'complex' TS (exhibiting multiple co-occurring conditions), but only 3% in other clusters of children with TS. Spencer et al. (1998)* found that language disorders were present in 6% of children with TS only and 20% of children with TS and co-occurring ADHD.

Receptive vocabulary was measured in five studies. Results all demonstrated average skills in children with TS (Bornstein et al., 1983; Ferrari et al., 1984; Jensen, 2004; Kalfayan Sweeten, 1997; Stokes et al., 1991). Jensen (2004) measured expressive vocabulary, reporting scores to be within the average range.

Pratt (2000) found no difference in confrontation naming accuracy between children with TS and a group of nondisabled peers, whereas Hulbert (1986) found that the mean accuracy score for a group of children with TS on confrontation naming was more than one SD below the mean of the norming sample. Four studies examined naming speed. Walenski et al. (2007)* measured naming speed and accuracy for manipulated objects (e.g., hammer) and non-manipulated objects (e.g., elephant). Children with TS and nondisabled peers were equal in accuracy for both naming tasks and were equal in speed for non-manipulated objects, but children with TS had faster naming speeds for manipulated objects. Jensen (2004) found that children with TS and children with TS and co-occurring dyslexia had average speeds for a rapid automatic naming (RAN) task. Schuerholz et al. (1996)* found no difference in RAN speeds between groups of children with TS, TS and co-occurring ADHD, and nondisabled peers. Schuerholz et al. (1998) again found no difference between RAN speeds for children with TS compared to nondisabled peers; but in contrast, children with TS and co-occurring ADHD were

significantly slower than children with TS only and nondisabled peers. Children with ADHD only were also slower than the group with TS only.

Ludlow et al. (1982)* measured language skills using a collection of language expression tasks (verbal fluency, sentence construction, digit repetition, reading and writing names) and receptive comprehension tasks (specific tasks not reported). They found that children with TS scored significantly lower than nondisabled peers on language expression tasks but not language comprehension tasks. How this pattern mapped to the specific tasks used was not further discussed. Hulbert (1986) found that children with TS scored, on average, more than one SD below the mean on a test of following directions. De Groot et al. (1997) found that children with TS plus co-occurring conditions (OCD and ADHD) had significantly more trouble with a categorisation task than other groups with TS. Verté et al. (2005) found that children with TS had stronger scores on a categorisation task compared to children with autism. Walenski et al. (2007)* studied the accuracy and speed of past tense production. They found that there was no difference between children with TS and nondisabled peers for accuracy, but children with TS completed the task faster than nondisabled peers for regular past tense words and for some irregular forms.

Children's ability to recall stories, repeat sentences or repeat non-words was measured in six studies. No difference between participants with TS and nondisabled peers was found for the accuracy of story recall (Channon et al., 2003b*; Pratt, 2000; Takács et al., 2018), sentence repetition (Hulbert, 1986; Ludlow et al., 1982) or non-word repetition (Dye et al., 2016*). Non-word repetition speed was significantly higher in participants with TS compared to nondisabled peers (Dye et al., 2016*).

Narrative language was evaluated using a fable task in a case series by Legg et al. (2005)*. This study performed a qualitative analysis of participant responses, concluding that 'disrupted language processing and reduced language usage' (p. 26) were present in 5/10 participants. Legg et al. (2005)* also evaluated specific areas of high-level language by administering the Test of Language Competence (subtests include Understanding Ambiguous Sentences, Making Inferences, Recreating Sentences and Understanding Metaphoric Expression). Half of the participants obtained scores below the average range. Of the five adolescents who experienced challenges with these tasks, four had co-occurring OCD, two had co-occurring ADHD, and one had no co-occurring conditions. Of those who did not experience challenges, all five had co-occurring OCD, one had co-occurring ADHD and one had co-occurring borderline personality disorder.

Anecdotal descriptions of language development in children with TS were reported in four studies that presented a series of cases. Kalfayan Sweeten (1997) found that language skills were developmentally appropriate in an adolescent male with TS. Of five cases studied by O'Quinn and Thompson (1980)*, three were reported to have language formulation problems, two were reported to have word-finding problems, and two were reported to have no language development problems. Thompson et al. (1979) reported on two children with TS, one with no language concerns, and one with language formulation, word-finding and coherence difficulties. Comings and Comings (1991) reported delayed communication skill development in a group of children with TS who also had autism. Fourteen of the 16 participants showed difficulties. Comings and Comings also evaluated three children with TS who had a family member with autism. No challenges with language development were noted in those three children.

Verbal IQ Findings

Twenty studies investigated the verbal intellectual abilities of children with TS using language-based subtests or composite scores on tests of intellectual function. Nineteen of these studies had not been discussed in previous reviews. Eighteen studies found that children with TS, on average, scored within the normal range of verbal intellectual ability (Bornstein, 1990; Bornstein et al., 1991; Carter et al., 2000; Cravedi et al., 2018; Ferrari et al.,1984; Huckeba et al., 2008; Hagin & Kugler, 1998; Hulbert, 1986; Incagnoli & Kane, 1983; Jensen, 2004; Kalfayan Sweeten, 1997; Khalifa et al., 2010; O'Quinn & Thompson, 1980; Shapiro et al., 1974, 1988; Stokes et al., 1991; Thompson et al., 1979; Yeates & Bornstein, 1994). Lanser et al. (1993) found that children with TS scored significantly lower for verbal IQ compared to children with right hemisphere dysfunction.

Some studies have looked at verbal IQ in relation to co-occurring diagnosis. Children with TS who had medium-to-low attention abilities (Huckeba et al., 2008) or co-occurring ADHD (Carter et al., 2000), scored lower on measures of verbal IQ compared to nondisabled peers, but children with high attention abilities scored similarly to nondisabled peers. On the other hand, Yeates and Bornstein (1994) found that the verbal IQ of children with both TS and ADHD was not significantly different from children with TS only. De Groot et al. (1997) compared verbal IQ in groups of children with TS only and children with TS and co-occurring conditions. Children with co-occurring OCD or both OCD and ADHD had significantly lower scores, whereas children with TS and ADHD scored similarly to those with TS only.

Verbal Fluency Findings

Verbal fluency is a test of lexical access based either on the first letter of words or on the semantic category (semantics) of words. Although often used as a measure of

neuropsychological function, verbal fluency is also strongly related to lexical ability (Whiteside et al., 2016). Fifteen studies presented results on the verbal fluency abilities of children with TS. The verbal fluency findings in five of these had not been previously reviewed. Letter fluency scores were reported in 10 studies. Six of these reported no differences between the letter fluency scores of children with TS and nondisabled peers (Channon et al., 2003b*; Church et al., 2009*; Mahone et al., 2001*, 2002*; Schuerholz et al., 1996, 1998); three reported average normative scores (Harris et al., 1995*; Hulbert, 1986; Jensen, 2004). Brookshire et al. (1994) found that children with TS scored lower than their unaffected siblings. The effect of having co-occurring conditions on letter fluency scores was considered in four studies.

Letter fluency scores of children with TS who also had ADHD did not differ from children with TS only (Channon et al., 2003b; Harris et al., 1995; Mahone et al., 2002), nondisabled peers (Channon et al., 2003b; Mahone et al., 2002; Schuerholz et al., 1996*, 1998*), children with TS who also had OCD (Channon et al., 2003b) or children with ADHD only (Harris et al., 1995; Mahone et al., 2002). Schuerholz et al. (1996)* and Schuerholz et al. (1998)* found that children who only had TS scored lower on letter fluency than children who had TS and ADHD. Mahone et al. (2001) compared children with ADHD only to children with TS only, finding no difference.

For category fluency, five studies reported scores for children with TS. No difference between the category fluency scores of children with TS and nondisabled peers were reported by Mahone et al. (2001)*, Mahone et al. (2002), Schuerholz et al. (1996)* and Schuerholz et al. (1998). Harris (1995)* found that, on average, children with TS scored within the normal range for their age but children with TS and ADHD scored lower than expected for their age. Brand et al. (2002)* found that children who also had ADHD performed more poorly than children with

TS only. Harris et al. (1995)* found no significant difference between performance for children with TS only, ADHD only and TS with co-occurring ADHD. Mahone et al. (2001) found that children with ADHD only performed more poorly than children with TS.

Four studies of verbal fluency did not differentiate between category and letter fluency. The scores of children with TS did not differ from nondisabled peers (Drury et al., 2018; Verté et al., 2005), children with TS and ADHD (Drury et al., 2018), children with TS and autism (Verté et al., 2005; described by authors as high functioning autism) or autism only (Verté et al., 2005). On the other hand, one study found that children with TS had higher verbal fluency scores than children with TS and OCD (regardless of whether or not they had ADHD; De Groot et al., 1997*). Drury et al. (2018) found that children with TS and ADHD had lower verbal fluency scores than nondisabled peers. Khalifa et al. (2010)* found that children with TS scored lower than would be expected based on age norms (scores were below the 25th percentile).

Findings Related to Social Communication

Findings related to social communication are presented here in four groupings, chosen according to the Adams (2005) taxonomy and adjusted based on the types of studies located. The groupings include, overall social communication, social interaction, social cognition and Autism traits. No studies were grouped under pragmatics (Adams' third category) and language processing studies are presented above (Adams' fourth category).

Six studies reported on the overall social communication abilities of children with TS using caregiver questionnaires. None of these findings had been previously presented in reviews of language in TS (although some related skills had been reported on—i.e., social interaction). Eapen et al. (2019), Güler et al. (2015) and Verté et al. (2005) found statistically significant increases in social communication problems for groups with TS compared to nondisabled peers.

The samples in these studies experienced co-occurring conditions representative of the TS population. Eapen et al. (2019) found that social communication was more of a challenge in a group with autism than in a group with TS. Darrow et al. (2017) found that children with TS plus OCD had stronger social communication scores compared to children with TS only and children with TS plus ADHD. Darrow et al. (2017) and Sukhodolsky et al. (2003) found no significant difference between children with TS and nondisabled peers, but found that children with TS and co-occurring ADHD had significantly more challenged social communication scores compared to nondisabled peers. Sukhodolsky et al. (2003) also studied children with ADHD only and found that they had more challenges compared to children with only TS but similar scores to children with TS plus ADHD. Darrow et al. (2017) found that the children with TS only and TS plus ADHD had more challenges than the children with TS plus OCD and ADHD. Dykens et al. (1990) found that social communication was not a particular weakness for children with TS relative to other intellectual, academic and adaptive skills measured. Conversely, Darrow et al. (2017) found that two groups with TS (TS with OCD and TS with both ADHD and OCD) had higher scores on the social communication measure compared to nondisabled peers.

Social interaction was measured in 13 studies. These studies measured either motivation to interact socially or skill in social interaction using caregiver questionnaires. Findings in 11 of these studies had never been reported in a previous review. Dykens et al. (1990) reported social interaction as a weakness for children with TS in relation to other adaptive skills. Eighty-five percent of Kadesjö and Gillberg's (2000) sample were reported to have significant problems interacting with same-age peers. Eapen et al. (2019) reported social interaction problems compared to nondisabled peers. Darrow et al. (2017)*, Gorman et al. (2010) and Güler et al. (2015)* found that children's scores were significantly poorer on measures of social interaction

compared to nondisabled peers. On the other hand, Bawden et al. (1998) found that when children with TS were compared to peers with a chronic condition (diabetes mellitus) there was no significant difference. Social interaction problems correlated significantly with problematic home behaviour, lowered quality of life and poorer family relations (Marek, 2006), as well as higher overall TS severity (Hulbert, 1986).

Three of these studies considered social interaction in children with TS and ADHD separately from children who had TS only. Carter et al. (2000) and Sukhodolsky et al. (2003) found that children with TS and co-occurring ADHD scored significantly lower on social interaction measures compared to children with only TS and compared to nondisabled peers. When comparing children with TS only and nondisabled peers, Carter et al. (2000) and Sukhodolsky et al. (2003) did not find significant differences in social interaction skills. Sukhodolsky et al. (2003) found that children with only TS had better social interaction scores than children with ADHD only, but the TS plus ADHD group scored similarly to the ADHD only group. Hoekstra et al. (2004) found that the presence of co-occurring ADHD predicted social insight problems but not social contact problems. Gallina (1989) compared social interaction in children with TS, children with ADHD and nondisabled peers. There was a significant difference among the groups with the ADHD group scoring lowest, the TS group scoring second lowest, and the group of nondisabled peers scoring highest. A post hoc analysis was not completed in order to determine which pairwise differences were significant.

Five studies examined skills of social cognition in children with TS. Only one new study (previously unreviewed) was added. Güler et al. (2015)* found that children with TS had poorer social cognition scores compared to nondisabled peers on a parent questionnaire. Drury et al. (2018)* also investigated comprehension of sarcasm that was presented in simulated scenarios.

The authors tested both direct sarcasm (e.g., remarking '[you're] the best cook in the world' in response to burned toast) and indirect sarcasm (e.g., remarking 'I'll hire you in my restaurant' in response to burned toast; examples from Drury et al. (2018), p. 491). When sarcasm was presented directly there was no difference across TS and nondisabled peers. When sarcasm was presented indirectly performance was significantly poorer in children with TS compared to nondisabled peers. Drury et al. (2018) also investigated children's ability to answer questions about mental states of people in these sarcastic scenarios. The TS group and nondisabled peers performed similarly on these questions; however, mental state identification was lower for children with TS who performed more poorly on the indirect sarcasm measure. Rajendran et al. (2005)* compared comprehension of sarcasm in children with TS to children with autism. No significant difference was found. Rajendran et al. (2005) also compared comprehension of figures of speech and recognition of inappropriate requests in children with TS to children with autism. Children with autism had significantly lower scores. A case series published by Baron-Cohen and Robertson (1995) used two false belief tasks and a deception task to evaluate whether or not six children (one with TS, two with autism and three with TS and autism) had a grasp of theory of mind. It was concluded that the child with TS had intact theory of mind, while those with autism (or TS and autism) did not. Drury et al. (2012)* found that children with TS could discriminate and name emotions in facial expressions and vocal prosody with similar accuracy to nondisabled peers. Compared to nondisabled peers, children with TS and ADHD had trouble recognising angry prosody and judging whether or not it was congruent with the message being said.

Six studies reported on autism traits in children with TS. Four of these had not been previously reviewed. Percentages of children in TS samples exhibiting autism traits were

reported by Darrow et al. (23%; 2017)*, Kadesjö and Gillberg (20%; 2000) and Wadman et al. (26%; 2016). The presence of autism traits was found to be significantly lower in groups of children with TS compared to children with autism (Eapen et al., 2019; Verté et al., 2005). When compared to a nondisabled group; however, children with TS scored significantly higher on measures of autism traits (Eapen et al., 2019; Güler et al., 2015*).

Discussion

This scoping review was undertaken with the purpose of locating existing studies about the language and social communication abilities of children with TS and describing the composition of that literature. The objectives were to identify the scope of the current literature, summarise the findings presented in studies, and identify gaps in this literature. In keeping with the purposes of a scoping review, we focussed on drawing conclusions about the state of the literature, rather than evaluating findings. Nonetheless, in some cases, study limitations were evident with regard to publication date, sample size, methodology and the tasks used to measure language and social communication skills.

Current State of the Literature

We found 56 records that measured skills of language or social communication in children with TS. The majority of these papers and books came from the fields of psychology/psychiatry. Only a single article, a case series of adolescents with TS, was published in a speech-language pathology-focussed journal (Legg et al., 2005, Clinical Linguistics).

Language

There was some variety in the skills measured across language literature, however, the majority of studies examined verbal IQ or verbal fluency, as we might expect when language is investigated from a psychology or psychiatry standpoint. We did not find any studies

investigating language ability at the level of conversation (i.e., using conversational language sample analysis). Also, no single study within this review provided a detailed description of the language system involving multiple domains (i.e., form, use and content) and modalities (i.e., expressive and receptive), using, for example, the omnibus tests common to speech-language pathology clinical practice. Rather, isolated aspects of language have frequently been reported within the context of questions about neuropsychological and social aspects of TS.

Social Communication

With regard to the scope of social communication skills measured across studies, social interaction has been measured most widely. Although there are several studies measuring social interaction, they have not observed the interaction skills of children with TS directly, looked at direct effects of tics on social interaction encounters, surveyed the points of view of the children themselves about their interactions or involved other familiar interactants such as peers. Measures of social communication were almost entirely composed of parent questionnaires and reported only a mean total score, giving little clue as to where reported functional communication challenges lie. Social cognition has received some research attention thus far as well, but several social cognitive skills remain unstudied. Rates of children with TS who exhibit traits of autism are well established in this literature.

Language and Social Communication in Children with TS: Summary of Findings

We summarise the findings of the literature with caution given the small number of studies that have examined language skills beyond verbal IQ tests and verbal fluency, and the fact that many of the reviewed studies have elements that limit interpretation or generalisation (e.g., case descriptions, studies lacking detailed reporting). On the whole, the reviewed literature points to verbal IQ and verbal fluency in TS as being consistent with typical expectations.

Single-word vocabulary appears to be an area of strength. The literature suggests elevated rates of language disorder among children with complex TS (i.e., with co-occurring diagnoses). There is preliminary evidence of high-level language challenges in some children with TS (Legg et al., 2005). There are also some very preliminary hints from case studies and more dated research that challenges exist in following directions, language formulation and word-finding (Hulbert, 1986; O'Quinn & Thompson, 1980; Thompson et al., 1979). When considering social communication, there is evidence for elevated presence of autism traits in children with TS, though lower than what is generally seen in autism. There is also evidence for difficulties in social communication, social interaction and social cognition, including understanding indirect sarcasm and recognising message congruence for an angry tone of voice (Drury et al., 2012, 2018).

Impact of Co-occurring Diagnoses

Whenever studies reported on the relationship between co-occurring diagnoses and outcomes, we have described those findings. Performance on various tasks was sometimes poorer for groups with co-occurring conditions. This pattern was seen in studies that investigated overall prevalence of language disorders, social interaction, recognising angry tone of voice, categorisation, naming speed and verbal IQ; however, it is important to note that not all studies showed this pattern and not all studies provided data on co-occurring conditions. Careful documentation of the presence of co-occurring conditions and their relationship to outcomes will be an important consideration in future language and communication research with children who have TS. Developing our understanding of language and social communication concerns that present in TS only (i.e., 'pure' TS) is of interest given past findings in social communication, social cognition and high-level language and given that we know problems can exist in areas of social, academic and cognitive development. On the other hand, since approximately 80% of

individuals with TS have co-occurring conditions (Martino & Leckman, 2013), future research should be careful about focusing exclusively on pure TS, as findings will represent only a small subset of the population. Further, findings may not represent the type of child who would most likely be referred to the SLP (i.e., the 'complex' child).

Refining and Extending Conclusions About Language and Social Communication

Consistent with previous reviews covering aspects of language and social communication in TS, this review has found that children with TS exhibit autism-like characteristics, speeded language processing and developmentally appropriate verbal fluency skills. High-level language challenges have also been identified; however, we emphasise that although these findings have repeatedly been presented in past reviews, they remain preliminary in nature.

When considering the literature on language in children with TS from a speech-language pathology perspective, we must be cautious about some conclusions that have been previously presented regarding expressive language, receptive language and language development more generally in TS. The terms 'expressive' and 'receptive' have been used inconsistently in past reviews on the subject and have sometimes been used to refer to other language-related skills. Some studies citing weak expressive language in children with TS have measured verbal fluency, speech sound perception or reading and writing (e.g., Brookshire, 1994; Bornstein et al., 1990; Ludlow, 1982; cited in de Nil, 2006, Eddy, 2021; Murphy & Eddy, 2013). Although verbal fluency represents an aspect of expressive language, the SLPs view of expressive language is much broader, usually involving a comprehensive look at elements of semantic, morphological, syntactic and pragmatic development, at the levels of the single-word, sentence and larger discourse units. Despite this, verbal fluency findings have been relied upon heavily in past conclusions about expressive language and overall language in TS (e.g., Channon et al., 2003b;

Mahone et al., 2001; cited by de Nil et al., 2006). Additionally, one review suggested that language comprehension concerns may exist secondary to social cognitive challenges in TS (Morand-Beaulieu et al., 2017b). While social cognitive challenges are very likely to have a complex effect on functional language use, we currently cannot rule out challenges in core receptive language skills in children with TS. With five studies demonstrating average receptive vocabulary, and two older studies showing challenges in following directions and performance on an unknown set of receptive language tasks (Bornstein et al., 1983; Ferrari et al., 1984; Hulbert, 1986; Jensen, 2004; Kalfayan Sweeten, 1997; Ludlow 1982; Stokes et al., 1991), we have barely scratched the surface of receptive language evaluation in this population.

Discussion of social communication in TS is mounting in the literature: several authors of past reviews have identified it as an area of concern in TS and literature on social cognitive challenges in adults has become well established (e.g., Eddy et al., 2010a, 2010b, 2011, 2016, 2017; Eddy & Cavanna, 2015). Some researchers are querying if social communication could be a central component of the disorder since individuals with TS often exhibit socially inappropriate behaviour and have challenges on tasks involving social reasoning and social interpretation (See Albin, 2018; Eddy, 2021; Eddy & Cavanna, 2013; Eddy et al., 2011). Only a few studies of social cognition in children exist currently and, as previously stated (e.g., Eddy et al., 2011; Morand-Beaulieu et al, 2017b), findings indicate that these challenges are subtle. Differences in social interpretation seem to appear in some contexts only (i.e., indirect sarcasm and understanding angry tone). The current review is the first to present the full range and depth of studies on social communication in children, and through employing the Adams (2005) taxonomy of social communication, has begun to organise this literature from a speech-language pathology point of view. This mapping indicates that social communication problems exist

across at least two of the four domains of social communication (Adams, 2005): social interaction and social cognition.

Although average or near average intellectual abilities are frequently discussed in TS literature, verbal IQ findings in children had not been reported previously with regard to language development. We identified this skill as a strength for children with TS after reviewing all of the available studies on the topic. Similarly, past reviews have not looked specifically at vocabulary, categorisation, or ability to recall/repeat language, all areas that we have identified as strengths.

Considerations for Future Research

Future reviews evaluating literature on language and social communication in TS should pay particular attention to study power, task difficulty and tool validity. A future review on this topic may choose to employ a systematic review methodology in order to formally evaluate the strength of evidence. To further broaden our knowledge-base, it would be beneficial for future studies to consider studies of children with other tic disorders, such as chronic motor vocal tic disorder and provisional tic disorder. Furthermore, the language abilities of adults with TS are also of interest and should be considered in future reviews.

From this mapping of the existing literature on language, we suggest that there is still a need for comprehensive study of language abilities in children with TS with a focus on coverage across domains and modalities and attention to high-level language abilities and discourse contexts. Larger sample sizes with control are needed to further preliminary findings in the literature. Additionally, there is a need for information about communication abilities in natural contexts where all of the factors that present for children with TS are at play (e.g., social perceptions, memory, tics). There is also a need for information about a larger range of social

cognitive skills in children, while keeping in mind the importance of differing task difficulty. Finally, future studies should focus on more detailed descriptions of social communication skills and social experiences, moving beyond measurement of autism-specific traits.

Potential role of the SLP in support of children with TS

Language development in children with TS has not been investigated in much detail in the speech-language pathology literature thus far. The summary provided here indicates that SLPs will be more likely to see children with complex forms of TS (e.g., those with TS and ADHD) on their caseloads. SLPs should pay attention to screening children with TS for language disorders, investigating social communication and social interaction development and investigating narrative language, language formulation and high-level language skills. Treatment strategies may involve directly targeting areas of challenge and/or providing appropriate accommodations (e.g., extra time to complete activities, modified activities, extra teacher instruction). As they get older, young people with TS may benefit from education and coaching to advance self-advocacy skills. Given the vast literature on neuropsychological development in children with TS, interdisciplinary collaboration could further clarify the role of the SLP in providing interventions for children experiencing challenges in their language and social communication development.

Limitations

When reporting conclusions about the current research findings, an important limitation to consider is that the quality of the evidence has not been systematically evaluated. Although the inclusion of research from prior to 2000 enabled a full review of existing literature, the age of some studies constrains the reader's ability to interpret findings as reporting standards and the definition and diagnostic process for TS have changed over time. Finally, this review focussed

on skills ‘within the child’, but it is important to recognise that contextual factors such as environment and social and self-stigma associated with tics may play an important role in the communication experiences of children with TS (e.g., Malli et al., 2016).

Conclusion

The present review revealed 56 studies investigating language and/or social communication abilities in children with TS. Although a large number of studies exist on this topic, some areas still remain under investigated. When considering the social and academic challenges that children with TS exhibit and the social cognitive challenges that adults with TS exhibit, we suggest that more comprehensive research into the language and communication skills of children with TS is needed. Collaboration with other disciplines, such as psychology, neuropsychology and psychiatry would be of benefit in future research and clinical endeavours as these disciplines have a well-established literature of research with this clinical population.

References

Adams, C. (2005). Social communication intervention for school aged children: rationale and description. *Seminars in Speech and Language, 26*(3), 282–288. https://doi.org/10.1055/s-2005-917123

Albin, R. (2018). Tourette syndrome: a disorder of the social decision making network. *Brain, 141*(2), 332–347.

American Psychiatric Association (APA). (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). APA. https://doi. org/10.1176/appi.books.9780890425596

Arksey, H., & O'Malley, L. (2005). Scoping studies: Towards a methodological framework. *International Journal of Social Research Methodology: Theory and Practice, 8*(1), 19–32. https://doi.org/10. 1080/1364557032000119616

Aromataris, E. & Riitano, D. (2014). Constructing a search strategy and searching for evidence. *American Journal of Nursing, 114*(5), 49–56. https://doi.org/10.1097/01.NAJ.0000446779.99522.f6

Baizabal-Carvallo, J., & Jankovic, J. (2022). Sex differences in patients with Tourette syndrome. *CNS Spectrums*, 1–7. https://doi.org/10. 1017/S1092852922000074

Baron-Cohen, S., Jolliffe, T., Mortimore, C. & Robertson, M. (1997). Another advanced test of theory of mind: Evidence from very high functioning adults with autism or Asperger syndrome. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 38*(7), 813–822.

Baron-Cohen, S. & Robertson, M.M. (1995). Children with either autism, Gilles de la Tourette syndrome or both: Mapping cognition to specific syndromes. *Neurocase, 1*(2), 101–104. https://doi.org/10. 1080/13554799508402353

Bawden, H. N., Stokes, A., Camfield, C. S., Camfield, P. R. & Salisbury, S. (1998). Peer relationship problems in children with Tourette's disorder or diabetes mellitus. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 39*(5), 663–668. https://doi.org/10. 1017/S0021963098002480

Bitsko, R. H., Claussen, A. H., Lichstein, J., Lichstein, J., Black, L. I., Jones, S. E., Danielson, M. L., Hoenig, J. M., Davis Jack, S. P., Brody, D. J., Gyawali, S., Maenner, M. J., Warner, M., Holland, K. M., Perou, R., Crosby, A. E., Blumberg, S. J., Avenevoli, S., Kaminski, J. W. & Ghandour, R. M. (2022). Mental health among children— United States, 2013–2019. *Morbidity and Mortality Weekly Report, 71*(2), 1–48.

Bloch, M. H. (2013). Clinical course and adult outcome in Tourette syndrome. In: D. Martino & J. F. Leckman (Eds.), *Tourette syndrome*, pp. 107–120. Oxford University Press. https://doi.org/10.1093/med/ 9780199796267.003.0005

Bloch, M. H., Sukhodolsky, D. G., Leckman, J. F. & Schultz, T. (2006). Fine-motor skill deficits in childhood predict adulthood tic severity and global psychosocial functioning in Tourette's syndrome. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 47*(6), 551–559. https://doi.org/10.1111/j.1469-7610.2005.0156 1.x

Bornstein, R.A. (1990). Neuropsychological performance in children with Tourette's syndrome. *Psychiatry Research, 33*(1), 73–81. https://doi.org/10.1016/0165-1781(90)90150-4

Bornstein, R. A., Baker, G. B., Bazylewich, T. & Douglass, A. B. (1991). Tourette syndrome and neuropsychological performance. *Acta Psychiatrica Scandinavica, 84*(3), 212–216. https://doi.org/10.1111/ j.1600-0447.1991.tb03131.x

Bornstein, R. A., King, G. & Carroll, A. (1983) Neuropsychological abnormalities in Gilles de la Tourette's syndrome. The Journal of Nervous and Mental Disease, 171(8), 497–502.

Brand, N., Geenen, R., Oudenhoven, M., Lindenborn, B., Van Der Ree, A., Cohen-Kettenis, P. & Buitelaar, J. K. (2002). Brief report: Cognitive functioning in children with Tourette's syndrome with and without comorbid ADHD. *Journal of Pediatric Psychology, 27*(2), 203–208. https://doi.org/10.1093/jpepsy/27.2.203

Brookshire, B. L., Butler, I. J., Ewing-cobbs, L., & Fletcher, J.M. (1994). Neuropsychological characteristics of children with Tourette syndrome: Evidence for a nonverbal learning disability. *Journal of Clinical and Experimental Neuropsychology, 16*(2), 289–302. https://doi.org/10.1080/01688639408402639

Burd, L., Christensen, T. & Kerbeshian, J. (2008). Speech, language, and communication in Tourette's syndrome. *Annual Review of Applied Linguistics, 28*, 170–190. https://doi.org/10.1017/ S026719050808001X

Burd, L., Freeman, R. D., Klug, M. G. & Kerbeshian, J. (2005). Tourette syndrome and learning disabilities. *BMC Pediatrics, 5*(34), 7–12. https://doi.org/10.1186/1471-2431-5-34

Carter, A. S., O'Donnell, D. A., Scahill, L., Schultz, R.T., Leckman, J. F. & Pauls, D. L. (2000). Social and emotional adjustment in children affected with Gilles de la Tourette's Syndrome: Associations with ADHD and family functioning. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 41*(2), 215–223. https://doi.org/10. 1017/S0021963099005156

Champion, L. M., Fulton, W. A. & Shady, G. A. (1988). Tourette syndrome and social functioning in a Canadian population. *Neuroscience and Biobehavioral Reviews, 12*(3–4), 255–257. https://doi. org/10.1016/S0149-7634(88)80054-X

Chang, S. W., McCracken, J. T. & Piacentini, J. C. (2007). Neurocognitive correlates of child obsessive compulsive disorder and Tourette syndrome. *Journal of Clinical and*

Experimental Neuropsychology, 29(7), 724–733. https://doi.org/10.1080/13825580600966383

Channon, S., Crawford, S., Vakili, K. & Robertson, M. M. (2003a). Real-life-type problem solving in Tourette syndrome. *Cognitive and Behavioral Neurology, 16*(1), 3–15. https://doi.org/10.1097/00146965-200303000-00002

Channon, S., Drury, H., Gafson, L., Stern, J. & Robertson, M. M. (2012). Judgements of social inappropriateness in adults with Tourette's syndrome. *Cognitive Neuropsychiatry, 17*(3), 246–261. https://doi. org/10.1080/13546805.2011.590689

Channon, S., Pratt, P. & Robertson, M. M. (2003b). Executive function, memory, and learning in Tourette's syndrome. *Neuropsychology, 17*(2), 247–254. https://doi.org/10.1037/0894-4105.17.2.247

Channon, S., Sinclair, E., Waller, D., Healey, L. & Robertson, M. M. (2004). Social cognition in Tourette's syndrome: Intact theory of mind and impaired inhibitory functioning. *Journal of Autism and Developmental Disorders, 34*(6), 669–677. https://doi.org/10.1007/ s10803-004-5287-x

Church, J. A., Wenger, K. K., Dosenbach, N. U. F., Miezin, F. M., Petersen, S. E. & Schlaggar, B. L. (2009). Task control signals in pediatric Tourette syndrome show evidence of immature and anomalous functional activity. *Frontiers in Human Neuroscience, 3*, 1–14. https://doi.org/10.3389/neuro.09.038.2009

Claussen, A.H., Bitsko, R.H., Holbrook, J.R., Bloomfield, J. & Giordano, K. (2018) Impact of Tourette syndrome on school measures in a nationally representative sample. Journal of Developmental and Behavioral Pediatrics, 39(4), 335–342. https://doi. org/10.1097/DBP.0000000000000550

Comings, D. E. & Comings, B. G. (1991). Clinical and genetic relationships between autism-pervasive developmental disorder and Tourette syndrome: A study of 19 cases. *American Journal of Medical Genetics, 39*(2), 180–191. https://doi.org/10.1002/ajmg. 1320390213

Cravedi, E., Deniau, E., Giannitelli, M., Pellerin, H., Czernecki, V., Priou, T., Xavier, J., Consoli, A., Hartmann, A. & Cohen, D. (2018). Disentangling Tourette syndrome heterogeneity through hierarchical ascendant clustering. *Developmental Medicine and Child Neurology, 60*(9), 942–950. https://doi.org/10.1111/dmcn.13 913

Darrow, S. M., Grados, M., Sandor, P., Hirschtritt, M. E., Illmann, C., Osiecki, L., Dion, Y., King, R., Pauls, D., Budman, C. L., Cath, D. C., Greenberg, E., Lyon, G. J., McMahon, W. M., Lee, P. C., Delucchi, K. L., Scharf, J.M. & Mathews, C. A. (2017). Autism spectrum symptoms in a Tourette's disorder sample. *Journal of the American Academy of Child and Adolescent Psychiatry, 56*(7), 610–617. https://doi.org/10.1016/j.jaac.2017.05.002

De Groot, C. M., Yeates, K. O., Baker, G. B. & Bornstein, R. A. (1997). Impaired neuropsychological functioning in Tourette's syndrome subjects with co-occurring obsessive-compulsive and attention deficit symptoms. *Journal of Neuropsychiatry and Clinical Neurosciences, 9*(2), 267–272. https://doi.org/10.1176/jnp.9.2.267

De Nil, L. F., Roth, I. D., Saltzman-Benaiah, J. & Sandor, P. (2006). Speech, language, and learning deficiencies associated with Tourette's syndrome. *Perspectives on Neurophysiology and Neurogenic Speech and Language Disorders, 16*(2), 12–16. https://doi.org/10.1044/nnsld16.2.12

Devinsky, O., Bear, D., Moya, K., & Benowitz, L. (1993). Perception of emotion in patients with Tourette's syndrome. *Neuropsychiatry, Neuropsychology and Behavioral Neurology, 6*(3), 166–169.

Donaher, J. (2006). Tourette's syndrome and stuttering. Perspectives on Fluency and Fluency Disorders, 16(2), 5–6. https://doi.org/10.1044/ ffd16.2.5

Donaher, J. (2008). *Speech fluency demonstrated by children with Tourette syndrome* (3320012) [Doctoral Dissertation, Temple University]. https://doi.org/10.1016/b978-012397720-5.50034-7

Drury, H., Channon, S., Barrett, R., Young, M. B., Stern, J. S., Simmons, H. & Crawford, S. (2012). Emotional processing and executive functioning in children and adults with Tourette's syndrome. *Child Neuropsychology, 18*(3), 281–298. https://doi.org/10.1080/09297049.2011.613811

Drury, H., Shah, S., Stern, J. S., Crawford, S. & Channon, S. (2018). Comprehension of direct and indirect sarcastic remarks in children and adolescents with Tourette's syndrome. *Child Neuropsychology, 24*(4), 490–509. https://doi.org/10.1080/09297049.2017.1284777

Dye, C. D., Walenski, M., Mostofsky, S. H. & Ullman, M. T. (2016). A verbal strength in children with Tourette syndrome? Evidence from a non-word repetition task. *Brain and Language, 160*, 61–70. https://doi.org/10.1016/j.bandl.2016.07.005

Dykens, E., Leckman, J., Riddle, M., Hardin, M., Schwartz, S. & Cohen, D. (1990). Intellectual, academic, and adaptive functioning of Tourette syndrome children with and without attention deficit disorder. *Journal of Abnormal Child Psychology, 18*(6), 607–615. https://doi.org/10.1007/BF01342750

Eapen, V., McPherson, S., Karlov, L., Nicholls, L., Črnčec, R. & Mulligan, A. (2019). Social communication deficits and restricted repetitive behavior symptoms in Tourette

syndrome. *Neuropsychiatric Disease and Treatment, 15*, 2151–2160. https://doi.org/10.2147/ ndt.s210227

Eddy, C. M. (2021). Tourette syndrome. In: Commings, L. (Ed.), *Handbook of pragmatic language disorders* (pp. 375–383). Springer.

Eddy, C. M. & Cavanna, A. E. (2013). Altered social cognition in Tourette syndrome: Nature and implications. *Behavioural Neurology, 27*(1), 15–22.

Eddy, C. M. & Cavanna, A. E. (2015). Triangles, tricks and tics: Hypermentalizing in response to animated shapes in Tourette syndrome. *Cortex, 71*, 68–75. https://doi.org/10.1016/j.cortex.2015.06.003

Eddy, C. M., Cavanna, A. E. & Hansen, P. C. (2017). Empathy and aversion: The neural signature of mentalizing in Tourette syndrome. *Psychological Medicine, 47*(3), 507–517.

Eddy, C. M., Macerollo, A., Martino, D. & Cavanna, A. E. (2015). Interpersonal reactivity differences in Tourette syndrome. *Psychiatry Research, 228*(3), 932–935. https://doi.org/10.1016/j.psychres.2015. 05.070

Eddy, C. M., Mitchell, I. J., Beck, S. R., Cavanna, A. E. & Rickards, H. E. (2010a). Altered attribution of intention in Tourette's syndrome. *Journal of Neuropsychiatry and Clinical Neurosciences, 22*(3), 348–351. https://doi.org/10.1176/jnp.2010.22.3.348

Eddy, C. M., Mitchell, I. J., Beck, S. R., Cavanna, A. E. & Rickards, H. E. (2010b). Impaired comprehension of nonliteral language in Tourette syndrome. *Cognitive and Behavioral Neurology, 23*(3), 178–184.

Eddy, C. M., Mitchell, I. J., Beck, S. R., Cavanna, A. E. & Rickards, H. (2011). Social reasoning in Tourette syndrome. *Cognitive Neuropsychiatry, 16*(4), 326–47.

Eddy, C. M., Cavanna, A. E., Rickards, H. E. & Hansen, P. C. (2016). Temporo-parietal dysfunction in Tourette syndrome: insights from an fMRI study of theory of mind. *Journal of Psychiatric Research, 81*, 102–11.

Ferrari, M., Matthews, W. S. & Barabas, G. (1984). Children with Tourette syndrome: Results of psychological tests given prior to drug treatment. *Developmental and Behavioural Pediatrics,* 5(3), 116–119. https://doi.org/10.1097/00004703-198406000-00002

Gallina, N. B. (1989). *Tourette's syndrome children: Significant achievement and social behavior variables* (Publication No. 9000024) [Doctoral dissertation, City University of New York]. ProQuest Dissertations and Theses Global.

Gorman, D. A., Thompson, N., Plessen, K. J., Robertson, M. M., Leckman, J. F. & Peterson, B. S. (2010). Psychosocial outcome and psychiatric comorbidity in older adolescents with Tourette syndrome: A controlled study. *British Journal of Psychiatry, 197*(1), 36–44. https://doi.org/10.1192/bjp.bp.109.071050

Güler, A. S., Berkem, M., Yazgan, Y. & Kalaça, S. (2015). Cognitive flexibility and social responsiveness in children and adolescents with Tourette syndrome. *Child Psychiatry and Human Development, 46*(6), 940–950. https://doi.org/10.1007/s10578-015-0533-3

Greene, D. J., Black, K. J., & Schlaggar, B. L. (2013). Neurobiology and Functional Anatomy of Tic Disorders. In: D. Martino & J. F. Leckman (Eds., pp. 238–275). *Tourette Syndrome*. Oxford University Press. https://doi. org/10.1093/med/9780199796267.003.0012

Hagin, R. A., & Kugler, J. (1998). School problems associated with Tourette syndrome. In: D. J. Cohen, R. D. Bruun, & J. F. Leckman (Eds.), *Tourette's syndrome and tic disorders: Clinical understanding and treatment* (pp. 223–236). John Wiley.

Harris, E. L., Schuerholz, L. J., Singer, H. S., Reader, M. J., Brown, J., Cox, C., Mohr, J., Chase, G. A. & Denckla, M. B. (1995). Executive function in children with Tourette Syndrome and/or Attention Deficit Hyperactivity Disorder. *Journal of the International Neuropsychological Society, 1*, 511–516. https://doi.org/10.1016/j.cortex. 2019.11.007

Hoekstra, P. J., Steenhuis, M. P., Troost, P. W., Korf, J., Kallenberg, C. G. M. & Minderaa, R. B. (2004). Relative contribution of attention deficit hyperactivity disorder, obsessive-compulsive disorder, and tic severity to social and behavioral problems in tic disorders. *Journal of Developmental and Behavioral Pediatrics, 25*(4), 272–279. https://doi.org/10.1097/00004703-200408000-00007

Huckeba, W., Chapieski, L., Hiscock, M. & Glaze, D. (2008). Arithmetic performance in children with Tourette syndrome: relative contribution of cognitive and attentional factors. *Journal of Clinical and Experimental Neuropsychology, 30*(4), 410–420. https://doi. org/10.1080/13803390701494970

Hulbert, T. (1986). *Neuropsychological subgrouping of Tourette children*. [Unpublished doctoral dissertation]. Wayne State University. https://doi.org/10.1017/CBO9781107415324.004

Hyter, Y. (2017). Pragmatic assessment and intervention in children. In: L. Cummins (Ed.), *Research in clinical pragmatics* (pp. 493–526). Springer International Publishing.

Incagnoli, T. & Kane, R. (1983). Developmental perspective of the Gilles de la Tourette syndrome. *Perceptual and Motor Skills, 57*, 1271–1281. https://doi.org/10.2466/pms.1983.57.3f.1271

Jensen, J. (2004). Neuropsychological and behavioural characteristics of children and adolescents with Tourette Syndrome compared to controls with dyslexia. Journal of *Allergy and Clinical Immunology, 130*(2), 556. https://doi.org/10.1016/j.jaci.2012.05.050

Kadesjö, B. & Gillberg, C. (2000). Tourette's disorder: Epidemiology and comorbidity in primary school children. *Journal of the American Academy of Child and Adolescent Psychiatry, 39*(5), 548–555. https://doi.org/10.1097/00004583-200005000-00007

Kalfayan Sweeten, K. J. (1997). *Developmental issues in male adolescents with Tourette's syndrome.* [Unpublished doctoral dissertation]. California School of Professional Psychology.

Khalifa, N., Dalan, M. & Rydell, A. M. (2010). Tourette syndrome in the general child population: Cognitive functioning and self-perception. *Nordic Journal of Psychiatry, 64*(1), 11–18. https://doi.org/ 10.3109/08039480903248096

Khalil, H., Peters, M., Godfrey, C.M., McInerney, P., Soares, C. B. & Parker, D. (2016). An evidence-based approach to scoping reviews. *Worldviews on Evidence-Based Nursing, 13*(2), 118–123. https://doi. org/10.1111/wvn.12144

Krah, B. (2002). Tourette syndrome: The anatomy of a tic: From the point of view of a person with TS. *ASHA Leader,* 7(14), 5–6. https:// doi.org/10.1044/leader.FTR2.07142002.5

Kwak, C. & Jankovic, J. (2002). Tourette syndrome: The neurology of a tic: From the point of view of the scientist. *ASHA Leader, 7*(14), 5–7. https://doi.org/10.1044/leader.FTR1.07142002.4

Lanser, J. B. K., Van Santen, W. H. C., Jennekens-Schinkel, A. & Roos, R. A. C. (1993). Tourette's syndrome and right hemisphere dysfunction. *British Journal of Psychiatry, 163*, 116–118. https://doi.org/10. 1192/bjp.163.1.116

Legg, C., Penn, C., Temlett, J. & Sonnenberg, B. (2005). Language skills of adolescents with Tourette Syndrome. *Clinical Linguistics and Phonetics, 19*(1), 15–33. https://doi.org/10.1080/ 02699200410001665270

Levac, D., Colquhoun, H. & O'Brien, K. K. (2010). Scoping studies: Advancing the methodology. *Implementation Science, 5*(1), 1–10. https://doi.org/10.1186/1748-5908-5-69.

Ludlow, C. L., Polinsky, R. J., Caine, E. D., Bassich, C. J. & Ebert, M. H. (1982). Language and speech abnormalities in Tourette syndrome. In A. J. Friedhoff & t. N. Chase (Eds.), *Gilles de la Tourette syndrome* (pp. 351–361). Raven Press.

Mahone, E. M., Cirino, P. T., Cutting, L. E., Cerrone, P. M., Hagelthorn, K. M., Hiemenz, J. R., Singer, H. S. & Denckla, M. B. (2002). Validity of the behavior rating inventory of executive function in children with ADHD and/or Tourette syndrome. *Archives of Clinical Neuropsychology, 17*(7), 643–662. https://doi.org/10.1016/S0887-6177(01)00168-8.

Mahone, E. M., Koth, C. W., Cutting, L., Singer, H. S. & Denckla, M. B. (2001). Executive function in fluency and recall measures among children with Tourette syndrome or ADHD. *Journal of the International Neuropsychological Society, 7*(1), 102–111. https://doi.org/ 10.1017/S1355617701711101

Malli, M. A., Forrester-Jones, R. & Murphy, G. (2016). Stigma in youth with Tourette's syndrome: A systematic review and synthesis. *European Child and Adolescent Psychiatry, 25*(2), 127–139. https://doi.org/10.1007/s00787-015-0761-x

Marek, E. (2006). *Social functioning and quality of life for individuals with Tourette's Syndrome.* [Unpublished Doctoral Dissertation]. The Chicago School of Professional Psychology. https://doi.org/10. 1016/j.jaci.2012.05.050

Martino, D. & Leckman, J. F. (2013). *Tourette syndrome*. Oxford University Press.

Morand-Beaulieu, S., Grot, S., Lavoie, J., Leclerc, J. B., Luck, D. & Lavoie, M. E. (2017a). The puzzling question of inhibitory control in Tourette syndrome: A meta-analysis. *Neuroscience and Biobehavioral Reviews, 80*, 240–262. https://doi.org/10.1016/j.neubiorev. 2017.05.006

Morand-Beaulieu, S., Leclerc, J. B., Valois, P., Lavoie, M. E., O'Connor, K. P. & Gauthier, B. (2017b). A review of the neuropsychological dimensions of Tourette syndrome. *Brain Sciences, 7*(8), 1–30. https://doi.org/10.3390/brainsci7080106

Murphy, T. & Eddy, C. M. (2013). Neuropsychological assessment in Tourette syndrome. In: D. Martino & J. F. Leckman (Eds.), *Tourette syndrome*. Oxford University Press, pp. 439–467.

O'Hare, D., Eapen, V., Helmes, E., McBain, K., Reece, J. & Grove, R. (2015). Factors impacting the quality of peer relationships of youth with Tourette's syndrome. *BMC Psychology, 3*(1), 1–13. https://doi. org/10.1186/S40359-015-0090-3

O'Quinn, A. N. & Thompson, R. J. (1980). Tourette's syndrome: An expanded view. *Pediatrics, 66*(3), 420–424. https://doi.org/10.1016/ s0002-7138(09)61008-6

Peters, M. D. J., Godfrey, C. M., Khalil, H. McInerney, P., Parker, D. & Soares, C. B. (2015). Guidance for conducting systematic scoping reviews. *International Journal of Evidence-Based Healthcare, 13*(3), 141–146.

Pratt, P. L. (2000*). Executive functioning and habit learning in children with Tourette Syndrome.* [Unpublished Doctoral Dissertation]. University College London.

Rajendran, G., Mitchell, P. & Rickards, H. (2005). How do individuals with Asperger syndrome respond to nonliteral language and inappropriate requests in computer-mediated communication? *Journal of Autism and Developmental Disorders, 35*(4), 429–443. https://doi.org/10.1007/s10803-005-5033-z

Schuerholz, L. J., Baumgardner, T. L., Singer, H. S., Reiss, A. L. & Denckla, M. B. (1996). Neuropsychological status of children with Tourette's syndrome with and without attention deficit hyperactivity disorder. *Neurology, 46*(4), 958–965. https://doi.org/10.1212/ WNL.46.4.958

Schuerholz, L. J., Singer, H. S. & Denckla, M. B. (1998). Gender study of neuropsychological and neuromotor function in children with Tourette syndrome with and without attention-deficit hyperactivity disorder. *Journal of Child Neurology, 13*(6), 277–282. https://doi.org/10.1177/088307389801300607

Shapiro, E., Shapiro, A. K. & Clarkin, J. (1974). Clinical psychological testing in Tourette's syndrome. *Journal of Personality Assessment, 38*(5), 464–478. https://doi.org/10.1080/00223891.1974.10120005

Shapiro, A. K., Shapiro, E., Young, J. G. & Feinberg, T. E. (1988). *Gilles de la Tourette syndrome* (2nd ed.). Raven Press.

Sherman, E. M. S., Shepard, L., Joschko, M. & Freeman, R. D. (1998). Sustained attention and impulsivity in children with Tourette syndrome: Comorbidity and confounds. *Journal of Clinical and Experimental Neuropsychology, 20*(5), 644–657. https://doi.org/10.1076/jcen.20.5.644.1118

Sikora, K., Roelofs, A., Hermans, D. & Knoors, H. (2019). Executive control in language production by children with and without language impairment. *International Journal of Language and Communication Disorders, 54*(4), 645–655. https://doi.org/10.1111/ 1460-6984.12470

Singer, H. S. (2013). The Neurochemistry of Tourette Syndrome. In: D. Martino & J. F. Leckman (Eds.), *Tourette syndrome* (pp. 276–300). Oxford University Press. https://doi.org/10.1093/med/9780199796267.003.0013

Spencer, T., Biederman, J., Harding, M., O'Donnell, D., Wilens, T., Faraone, S., Coffey, B., & Geller, D. (1998). Disentangling the overlap between Tourette's disorder and ADHD. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 39*(7), 1037–1044. https://doi.org/10.1017/S0021963098002984

Stokes, A., Bawden, H. N., Camfield, P. R., Backman, J. E. & Dooley, M. (1991). Peer problems in Tourette's disorder. *Pediatrics, 87*(6), 936– 942.

Sukhodolsky, D. G., Scahill, L., Zhang, H., Peterson, B. S., King, R. A., Lombroso, P. J., Katsovich, L., Findley, D. & Leckman, J. F. (2003). Disruptive behavior in children with Tourette's syndrome: Association with ADHD comorbidity, tic severity, and functional impairment. *Journal of the American Academy of Child and Adolescent Psychiatry, 42*(1), 98–105. https://doi.org/10.1097/00004583- 200301000-00016

Takács, Á., Kóbor, A., Chezan, J., Éltető, N., Tárnok, Z., Nemeth, D., Ullman, M. T., & Janacsek, K. (2018). Is procedural memory enhanced in Tourette syndrome? Evidence from a sequence learning task. *Cortex, 100*, 84–94. https://doi.org/10.1016/j.cortex. 2017.08.037

Thompson, R. J., O'Quinn, A.N. & Logue, P. E. (1979). Gilles de la Tourette syndrome: a review and neuropsychological aspects of four cases. *Journal of Pediatric Psychology, 74*(4), 371–387. https:// doi.org/10.1177/014107688607900508

Tricco, A. C., Lillie, E., Zarin, W., O'Brien, K. K., Colquhoun, H., Levac, D., Moher, D., Peters, M. D. J., Horsley, T., Weeks, L., Hempel, S., Akl, E. A., Chang, C., McGowan, J., Stewart, L., Hartling, L., Aldcroft, A., Wilson, M.G., Garritty & Straus, S. E. (2018). PRISMA extension for scoping reviews (PRISMA-ScR): checklist and explanation. *Annals of Internal Medicine, 169*(7), 467–473. https://doi.org/10.7326/M18-0850

Verté, S., Geurts, H. M., Roeyers, H., Oosterlaan, J. & Sergeant, J. A. (2005). Executive functioning in children with autism and Tourette syndrome. *Development and Psychopathology, 17*(2), 415– 445. https://doi.org/10.1017/S0954579405050200

Wadman, R., Glazebrook, C., Beer, C. & Jackson, G. M. (2016). Difficulties experienced by young people with Tourette syndrome in secondary school: A mixed methods description of self, parent and staff perspectives. *BMC Psychiatry, 16*(1), 1–11. https://doi.org/10.1186/s12888-016-0717-9

Walenski, M., Mostofsky, S. H. & Ullman, M. T. (2007). Speeded processing of grammar and tool knowledge in Tourette's syndrome. *Neuropsychologia, 45*(11), 2447–2460. https://doi.org/10. 1016/j.neuropsychologia.2007.04.001

Yeates, K. O. & Bornstein, R. A. (1994). Attention deficit disorder and neuropsychological functioning in children with Tourette's syndrome. *Neuropsychology, 8*, 65–74. https://doi.org/10.1037/0894- 4105.8.1.65

Zhu, Y., Leung, K. M., Liu, P. Z., Zhou, M., & Su, L. Y. (2006). Comorbid behavioural problems in Tourette's syndrome are positively correlated with the severity of tic

symptoms. *Australian and New Zealand Journal of Psychiatry, 40*(1), 67–73. https://doi.org/10.1111/ j.1440-1614.2006.01745.x

Chapter 3: Communication and Psychosocial Functioning in Children with Tourette Syndrome: Parent-Reported Measures (Study 2)

Abstract

Background. Prior research suggests that some children with Tourette Syndrome experience communication challenges. However, little is known about the nature of challenges in children who do have these difficulties.

Aims. This study aimed to describe the proportion of children in a North American sample who present with challenges in communication development based on a standardized parent questionnaire and to determine if communication skills differ from normative samples, if parent reports of co-occurring conditions relate to these skills, and if these skills are related to parent-reported psychosocial functioning.

Methods & Procedures. A series of questionnaires were disseminated to parents in North America via Tourette Syndrome parent groups on social media (US and Canada), Tourette Syndrome associations (US and Canada), and medical clinics serving Tourette Syndrome populations (Canada). We collected demographic information, background information about tics and co-occurring conditions, information about communication using the Children's Communication Checklist, Second Edition (CCC-2), and information about psychosocial functioning using the Strengths and Difficulties Questionnaire (SDQ).

Outcomes & Results. CCC-2 results found that 62% of children had age-expected communication skills and 38% had challenges in communication development. Communication skills were significantly lower compared to the CCC-2 US population-based normative sample in

general communication, social-pragmatic communication, semantics, coherence, initiation, scripted language, context, nonverbal communication, social relations, and interests. There was a negative correlation between communication and psychosocial measures: as general communication and social-pragmatic communication skills decreased, psychosocial challenges increased ($r = -.44$, $p < .001$). The presence of co-occurring conditions did not predict general communication or social-pragmatic communication challenges.

Conclusions & Implications. Speech-language pathologists (SLPs) can expect most children with TS to have age-appropriate communication development, however, many will have challenges in at least one area of communication. Medical professionals should refer children to the SLP as early as possible if they suspect communication challenges, paying particular attention to children with higher tic disorder severities. SLPs should assess both core language and social aspects of communication development when children are referred and interventions should support communicative function while also encouraging acceptance of TS and the social differences that coexist with tics and TS.

Introduction

Tourette syndrome (TS) is a neurodevelopmental condition characterized by motor and vocal tics that appear during childhood (American Psychological Association, 2013). About 80% of people with TS have other co-occurring disorders, such as attention-deficit/ hyperactivity disorder (ADHD), obsessive-compulsive disorder (OCD) and/or anxiety (Cath et al., 2022).

Language disorders are thought to occur at a rate of between 12-30% in children with TS, higher than we would expect in the general population (10%; Norbury et al., 2016). Thus, some children with TS have co-occurring language disorders, but many do not. In fact, past research points to many strengths in language development for children with TS. Recent research has largely focused on language processing at the single-word level and has demonstrated strengths in verbal fluency and single-word vocabulary (see Feehan & Charest, 2023). Certain elements of syntactic processing appear to be a strength in children with TS as well; they can complete some grammatical tasks more quickly than typically developing peers while maintaining an equal level of accuracy (Walenski et al., 2007).

For those children who do experience communication challenges (defined as challenges in language and/or social communication), there is little literature detailing where areas of need might be. There are a few very dated case studies suggesting that language formulation, coherence, and word-finding could be areas of challenge for children with TS (O'Quinn & Thompson, 1980; Thompson et al., 1979). Further, a larger but similarly outdated study suggests following directions may be a challenge (Hulbert, 1986). There are also a few studies that suggest challenges with fluent expressive language formulation (de Nil et al., 2005; Donaher, 2008). A more recent case study identified challenges in high-level language (i.e., inferencing and non-literal language comprehension) and narrative language (Legg et al., 2005).

Autistic characteristics and social communication skills have been measured using the Autism Spectrum Screening Questionnaire (Ehlers et al., 1999), the Social Communication Questionnaire (Rutter et al., 2003), the Social Responsiveness Scale (Constantino & Gruber, 2012), and the first edition of the Children's Communication Checklist (Bishop, 1989; Darrow et al., 2017; Eapen et al., 2019; Güler et al., 2015; Kadesjö & Gillberg, 2000; Verté et al., 2005; Wadman et al., 2016). These studies have identified a high prevalence of autistic characteristics and/or challenges in social communication aspects of these tools. However, we currently have little detail about the nature of social communication challenges. Despite recent suggestions that social communication challenges could be a central component of TS (Albin, 2018; Eddy, 2021; Eddy & Cavanna, 2013; Eddy et al., 2011), the nature of social communication concerns in children with TS is not well understood. Further, people who live with tics in our social world experience societal stigma and differences in their social experiences (Suh et al., 2022). The relationship between these factors and social communication differences in TS has also not been explored.

Some research has addressed how co-occurring conditions might contribute to language and social communication skills in children with TS, but few studies have compared language skills in children with and without co-occurring conditions. Two studies reported that rates of language delays/disorders are high in children with co-occurring conditions (20-45%) and lower in children with "pure" TS (3-6%; Cravedi et al., 2018; Spencer et al., 1998). De Groot et al. (1997) found that children with Attention-Deficit/ Hyperactivity Disorder (ADHD) and/or Obsessive-Compulsive Disorder (OCD) performed more poorly than children with TS-only on a categorization test that measured concept formation (i.e., semantics). Similarly, Sukhodolsky (2003) found that children with TS-only had similar expressive and receptive language skills as

their peers, but children with co-occurring ADHD had significantly lower skills compared to their peers. A few studies have compared social communication skills in children with and without co-occurring conditions. A few studies reported that children with co-occurring ADHD perform similarly to children with TS-only on "social communication" measures (which look at a broad range of social-pragmatic language skills), but have lower scores on the "social interaction" measures (which specifically measure skills and motivation for interactions in social situations) (Carter et al., 2000; Darrow et al; 2017 & Sukhodolsky et al., 2003). Conversely, Pringsheim & Hammer (2013) found that a co-occurring diagnosis of ADHD contributed significantly to lower social communication scores in children with a range of tic disorders, including TS. Another study found that children with co-occurring OCD had stronger social communication scores compared to some of the other groups evaluated (i.e., TS-only and TS with ADHD; Darrow et al., 2017).

More generally, ADHD, OCD, and anxiety in children have all been associated with some specific communication challenges. Children with ADHD tend to have problems in pragmatics (social communication), high-level language/discourse, and language formulation (Helland et al., 2014; Korrel et al., 2017; Vaïsänen et al., 2014; Redmond et al., 2004). While adolescents with OCD are reported to have strong conversational skills, peer skills, and speech-act use, nonverbal communication and several other pragmatic language skills were found to be lower compared to nondisabled peers (Ghahari et al., 2017). Towbin et al. (2005) reported that children with mood and anxiety disorders generally scored within the average range on the communication composite of the Children's Communication Checklist, Second Edition (CCC-2; Bishop, 2003); however, 32% of children had low social interaction composite scores. Van

Steensel et al. (2013) found that children with anxiety disorders have elevated levels of communication challenges compared to nondisabled peers.

Psychosocial functioning considers aspects of social and psychological function and children with TS have been found to have challenges in both areas (e.g., Gutierrez-Colina et al., 2015). Psychosocial challenges may relate to a wide range of factors including co-occurring conditions (such as ADHD; OCD; and anxiety), severity of tics, school experiences, parenting styles and caregiver burden, motor skills, the complexities of living with tics in a social world, and other socio-cultural factors (Robertson & Eapen, 2017). The correlation between communication development and overall psychosocial skills has not been investigated in previous research.

In summary, there is some evidence that communication challenges are common in children with TS; however, we have little information about what these challenges look like and what other factors may be related. The purpose of this research was to investigate the extent to which a North American sample of children with TS experience communication challenges, to explore how parents rate their children in various communication domains, to understand how co-occurring conditions contribute to these skills, and to address the gap in our understanding of how these skills may relate to psychosocial skills. Communication skills were measured using the CCC-2 and psychosocial skills were measured using the Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997).

The following research questions were addressed:

1. What proportion of children with TS demonstrate parent-reported challenges in General Communication and Social-Pragmatic Communication skills on the CCC-2, and do these skills differ from a normative sample? What proportion of

children with parent-reported challenges had an existing communication disorder diagnosis? We expected that both mean General Communication and mean Social-Pragmatic Communication composite scores would be lower for our TS sample compared to children in the general population. Based on previous literature, we expected means similar to the norm for speech, syntax, and semantics. For all other domains of the CCC-2, we expected domain scores below norms since each domain is associated with coherence, high-level language, social communication, and/or autistic characteristics, all areas that past evidence indicates may be weaknesses.

2. How do co-occurring conditions predict General Communication and Social-Pragmatic Communication Composites in children with TS? We expected that the presence of ADHD, OCD and Anxiety would be associated with lower scores on the General Communication and Social-Pragmatic Communication Composites.
3. What is the relationship between communication functioning and levels of parent-reported psychosocial functioning in children with TS? We expected a negative relationship between scores on the Communication Composites (General and Social-Pragmatic) and the SDQ difficulties score.

Methods

Questionnaire Structure and Development

A series of questionnaires was created in REDCap consisting of four sections: background questions, communication questions (CCC-2), tic questions (Global Tic Rating Scale (GTRS; Gadow & Paolocelli, 1986), and psychosocial questions (SDQ). Section one was a list of background questions developed collaboratively by three of the authors. Background questions

collected information about who diagnosed the child with TS, age at diagnosis, current age, sex and gender, country and region, co-occurring conditions, languages spoken in the home, parent education, and race/ethnicity. A full list of background questions is provided in Appendix C.

Section two evaluated the communication skills of children using the CCC-2 (license No. LSR–262494). The CCC-2 is a 10-domain parent questionnaire for children from 4-16 years that asks parents to rate 70 questions on a four-point scale where 0 = less than once a week (or never); 1 = at least once a week, but not every day (or occasionally); 2 = once or twice a day (or frequently); and 3 = several times (more than twice) a day (or always). Age-based norms are available for each domain (M = 10, SD = 3) and for the General Communication Composite (M = 100, SD = 15). The General Communication Composite has demonstrated reliability and validity and a cut score of 85 provides good sensitivity and specificity for differentiating between children with language disorders and typical language development. The CCC-2 was selected as the communication measure because it has been used extensively in past research both for identifying childhood language disorders and for understanding social communication profiles in groups where social communication skills are of interest, such as autism, traumatic brain injury, ADHD, emotional-behavioural disorders, Williams syndrome, and anxiety (Bignell & Cain, 2007; Fisher et al ., 2022; Mackie & Law, 2010; Philofsky et al., 2007; Towbin et al., 2005). The overall communication skills of participants were measured using the General Communication Composite of the CCC-2, which combines eight domains: Speech, Syntax, Semantics, Coherence, Initiation, Scripted Language, Context, and Nonverbal Communication. Social communication was measured by creating a Social-Pragmatic Communication Composite, which combines Initiation, Context, and Nonverbal Communication domains (which are also included in the General Communication Composite). This combination is not normed in the CCC-2 test

manual but has been used to measure social communication on the CCC-2 in past research (Saul et al., 2022). This composite aligns well with our intention to understand social communication because it captures social-pragmatic language skills (i.e., social communication skills) independently from form/content language skills (i.e., language structure, vocabulary, and discourse).

The skills measured by individual domains of the CCC-2 focussed on the following aspects of communication:

A. *Speech*: Articulation errors, intelligibility and fluency of speech.
B. *Syntax*: Sentence length and complexity and correct use of verbs, pronouns, and morphemes.
C. *Semantics*: word choice and word retrieval.
D. *Coherence*: Ability to provide logical explanations/descriptions, use clear referents, provide context, and sequence ideas.
E. *Initiation*: Consideration of audience and timing when deciding to talk to others. Questions focus on detecting children who initiate at a higher frequency than what is typical.
F. *Scripted Language*: Use of memorized or overly precise intonation or word choice, echolalia, and communication partner's enjoyment of conversation.
G. *Context*: Adjustment of communication style to the needs of others, considering all information available when communicating, and comprehension of jokes, puns, and non-literal language.
H. *Nonverbal Communication*: Use and comprehension of facial expression, proxemics, and use of gestures and eye contact.
I. *Social Relations*: Social anxiety, social responsiveness, and social acceptance.

J. *Interests*: Preference for unique/specific activities and conversation topics, rote communication style, and preference for predictability.

Section three of the survey collected information about the severity of TS using the GTRS (use agreement date: November 21, 2022). There are four severity questions rated on a four-point scale about how noticeable the tics are to others, how embarrassing they are for the child, how much they affect school and home functioning, and to what degree they cause social rejection (0-1 = low; 2 = medium; 3 = high). The GTRS was selected because it is a five-minute measure that can categorize children into three groups based on the severity of their tic disorder. It was "suggested" as a measure by experts in the field, but has not been psychometrically evaluated (Martino et al., 2017). Measures of this type that have psychometric information available are much longer (>20 minutes) or need to be completed by a clinician.

Section four collected information about psychosocial functioning using the SDQ (authorization invoice No. 101811). The SDQ is a measure for children 3-16 years of age. It yields a total score for psychosocial difficulties using items focused on emotional symptoms, conduct problems, hyperactivity/inattention, and peer relationship problems. Total scores are broken into four categories (0-13 = 'close to average;' 14-16 = 'slightly raised;' 17-19 = 'high;' 20-40 = 'very high'). The SDQ has demonstrated validity and reliability for evaluating psychosocial functioning.

The REDCap version of the questionnaires was piloted in two phases. The first phase involved sending the survey to four professionals with research and/or clinical training in communication sciences to seek feedback and corrections. After the suggested changes were made, the second phase involved sending the survey to three parents of children within the 8-16 years range who had communication and/or neurodevelopmental concerns (other than TS) to

seek feedback and corrections. The updated survey represents the final version sent to participants.

Inclusion Criteria and Survey Dissemination

Parents of children with TS between the ages of 8-16 were recruited through social media and regional TS organizations across North America and through local medical clinics that provide health services to people with TS. They were invited to complete the survey online. Potential participants were provided with a link and, upon clicking, were directed to an information letter and a statement of implied consent. Participants were able to access the survey if they agreed to a copyright statement indicating that they would not copy the content of questionnaires and if their answers to the five eligibility questions indicated that they fit the eligibility criteria (questions about age, diagnosis, and geographic location). Children were included if they were located in North America and did not have a diagnosis of autism or intellectual disability. The survey was started by 156 respondents and 64 of them submitted the survey. Each submitted response was checked for completion. Three surveys we excluded due to missing responses, leaving 61 surveys. The survey was open from Nov 2, 2022 to Nov. 9, 2023. This research was reviewed by the Human Research Ethics Board at [Name of university redacted for review], Pro00126653.

The children of the respondents were 41 males and 20 females, 58 children were cisgender, 2 were transgender, and one did not specify. Children were 8-16.5 years of age (M = 12.5 years). Parents reported that children had been diagnosed with TS by their family doctor (n = 3), pediatrician (n = 10), psychiatrist (n = 18), neurologist (n = 25), or did not specify (n = 5). Age at diagnosis ranged from 3-14 years (M = 8.2 years). The racial/ethnic backgrounds reported by participants included Japanese (n = 1), Jewish (n = 1), White (n = 51), White/Filipino (n = 2),

White/Black (*n* = 1), White/Indigenous (*n* = 1), White/Latin American (*n* = 1), White/South Asian (*n* = 1), and White/Southeast Asian (*n* = 1) (missing data *n* = 1). Primary languages spoken in the home included English (*n* = 57) and French (*n* = 4). Nine households reported being multilingual (Arabic, Bengali, French, Spanish, and Ukrainian). All parent respondents had completed high school and 56 (92%) had received college or university education. Tables 2 and 5 include participant geographic locations, co-occurring conditions, and tic disorder severity as determined by the GTRS. Thirty-one had low tic disorder severity (51%), 20 had a medium tic disorder severity (33%), and 10 had a high tic disorder severity (16%).

Table 2

Participant Location

Location (*n*=61)	*n*(%)
Canada	43(70)
Atlantic Canada	4(7)
Eastern Canada	15(25)
Western Canada	19(31)
Prairies	5(8)
United States	18(30)
Midwest	3(5)
Northeast	10(16)
Southeast	1(2)
Southwest	2(3)
West	2(3)

Results

Analysis

Scoring of the CCC-2 and SDQ were conducted according to the instructions of test developers using paper forms. Totals from paper forms were transferred to an Excel spreadsheet along with REDCap data outputs. Data were checked for normality, independence of errors, homoscedasticity, outliers, and multicollinearity in SPSS. To determine whether or not language and social communication scores differed from a normative sample (research question one), we used one-way single sample t-tests to compare General Communication Composite, Social-Pragmatic Communication Composite (combining Initiation, Context and Nonverbal Communication domains), and individual domain scores that passed tests of normality (Coherence and Scripted Language) to a normative sample. Speech, Syntax, Semantics, Social Relations, and Interests domains did not pass tests of normality; therefore, we used the Wilcoxon one sample signed ranks test. We used the Holm-Bonferroni method to control for family-wise error ($p = .05/9 = .006$). We used two regression models to test the contribution of co-occurring conditions to General Communication and Social-Pragmatic Communication Composites (research question two). We used linear regression to determine if there was a negative relationship between each communication composite score and psychosocial functioning (research question three).

Language and Social Communication Skills (Research Question 1)

Table 3 lists the means, standard deviations, and ranges for scores on composites and individual domains of the CCC-2. Sixty-nine percent of participants ($n = 42$) received a General Communication Composite score within one SD of the mean (or higher) and 31% ($n = 19$) received a score more than one SD below the mean. Sixty-six percent of participants ($n = 40$)

received a Social-Pragmatic Communication score within one SD of the mean (or higher) and 34% (*n* = 21) received a score more than one SD below the mean. Sixty-two percent of participants (*n* = 38) received scores within one SD of the mean (or higher) for *both* composites, 28% (*n* = 17) received scores more than one SD below the mean for *both* composites, and the total number of children with low General Communication *and/or* Social-Pragmatic Communication scores was 23 (38%). The General Communication and Social-Pragmatic Communication scores for children with and without a communication disorder diagnosis are presented in Table 4. Numbers of participants with scores more than one SD below the mean for individual domains were as follows: *Speech* = 9 (15%), *Syntax* = 7 (11%), *Semantics* = 12 (20%), *Coherence* = 18 (30%), ***Initiation*** = 22 (36%), *Scripted Language* = 12 (20%), ***Context*** = 11 (18%), ***Nonverbal Communication*** = 19 (31%), Social Relations = 26 (43%), Interests = 23 (38%). Italicized domains are included in the General Communication Composite and bold italicized domains are included in the Social-Pragmatic Communication Composite.

Mean/median performance on the CCC-2 General Communication Composite, Social-Pragmatic Communication Composite, Semantics, Coherence, Scripted Language, Social Relations, and Interests were significantly lower than the population mean/median as a whole. All effect sizes were medium to large. Speech and Syntax were not significantly different from the population median.

Table 3

Language and Social Communication Test Score Descriptive Statistics (CCC-2)

Score	Minimum	Maximum	Mean (SD)	Test statistic ($df = 60$)	Effect size (d)
General Communication Composite (Normative sample $M = 100$, $SD = 15$)	60	117	89.8(13.5)*	$t = -5.90$	0 .76
Social-Pragmatic Composite[a] (Normative sample $M = 10$, $SD = 3$)	2	13	7.8(2.4)*	$t = -7.45$	0.95
Domain subscale (Normative sample $M = 10$, $SD = 3$)					
Speech	1	12	9(11)[b]	$z = -1.30^{n.s.}$	0.17
Syntax	4	12	9(8)[b]	$z = -.13^{n.s.}$	0.02
Semantics	2	13	8(11)[b]	$z = -3.81$*	0.49
Coherence	3	13	8.0(2.8)	$t = -5.65$*	0.72
Initiation	1	15	7.6(2.7)[a]		
Scripted Language	3	13	8(9)[b]	$z = -4.74$*	0.61
Context	2	13	8.3(2.6)[a]		
Nonverbal Communication	1	12	7.3(2.9)[a]		
Social Relations	1	12	7(11)[b]	$z = -6.32$*	0.81
Interests	1	16	7(15)[b]	$z = -4.67$*	0.60

[a]The Social-Pragmatic Communication Composite was composed of Initiation, Context, and Nonverbal Communication; therefore,

individual domain significance was not tested.

[b]Median (range) are reported because assumptions of normality were violated

* $p < .001$

[n.s.] Not Significant

Note: Italicized domains are included in the General Communication Composite and bold italicized domains are included in the Social-Pragmatic Communication Composite.

Table 4

General Communication and Social-Pragmatic Communication Scores for Children With and Without a Communication Disorder Diagnosis

		Scores <2SD below mean *n*(%)	Scores 1SD-2SD below mean *n*(%)	Scores within the mean +/-1SD *n*(%)
Diagnosed with CD *n*=6	General Communication Composite	4(67)	2(33)	0(0)
	Social-Pragmatic Communication Composite	2(33)	4(67)	0(0)
Not diagnosed with CD *n*=55	General Communication Composite	2(4)	11(20)	42(76)
	Social-Pragmatic Communication Composite	2(4)	13(24)	40(73)

CD = Communication Disorder

The Contribution of Co-occurring Conditions to General Communication and Social-Pragmatic Communication Skills (Research Question 2)

The numbers of participants with each co-occurring condition are presented in Table 5. Nearly all participants reported at least one co-occurring condition (n = 60, 98%) and fifty-four children reported two or more co-occurring conditions (89%). Only one participant had received a diagnosis of TS-only (2%). The regression model testing ADHD, OCD and anxiety as predictors of General Communication was not significant $F(3,57) = 1.11$, $p = .23$. The model explains 2.4% of the variance of General Communication Composite Standard Score (adjusted $R^2 = .024$). None of the predictors accounted for a significant amount of unique variance.

Similarly, the model testing ADHD, OCD and anxiety as predictors of Social-Pragmatic Communication was not significant $F(3,57) = 2.11$, $p = .11$. The model explains 5.3% of the variance of General Communication Composite Standard Score (adjusted $R^2 = .053$). None of the predictors accounted for significant unique variance. Figures 4 and 5 include box and whisker plots for General Communication and Social-pragmatic Communication for children with and without each diagnosis.

Table 5

Parent-Reported Co-occurring Conditions and GTRS Tic Disorder Severity

Condition (confirmed or suspected) (*n*=61)	*n*(%)[a]	Tic Disorder Severity		
		Low *n*=31	Medium *n*=20	High *n*=10
Language/communication disorder	7(11)	2	1	4
Speech disorder	9(15)	3	2	4
Hearing loss	0(0)	0	0	0
ADHD	45(74)	24	13	8
OCD	33(54)	15	9	9
LD	15(25)	6	5	4
Anxiety	48(79)	24	15	9
Depression	16(26)	7	8	1

GTRS - Global Tic Rating Scale; ADHD – Attention-Deficit/ Hyperactivity Disorder; OCD-Obsessive-Compulsive Disorder; LD - Learning Disability

[a]Totals > 100% because respondents could select more than one option.

Figure 4

Box and Whisker Plots for General Communication for Children With and Without ADHD, OCD and Anxiety

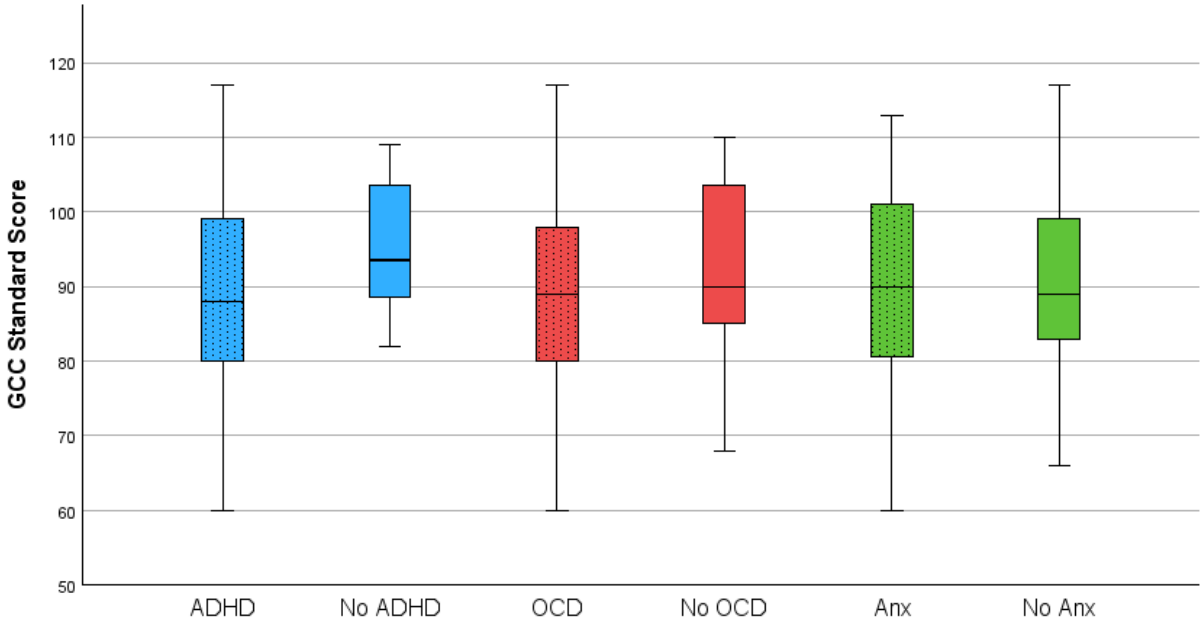
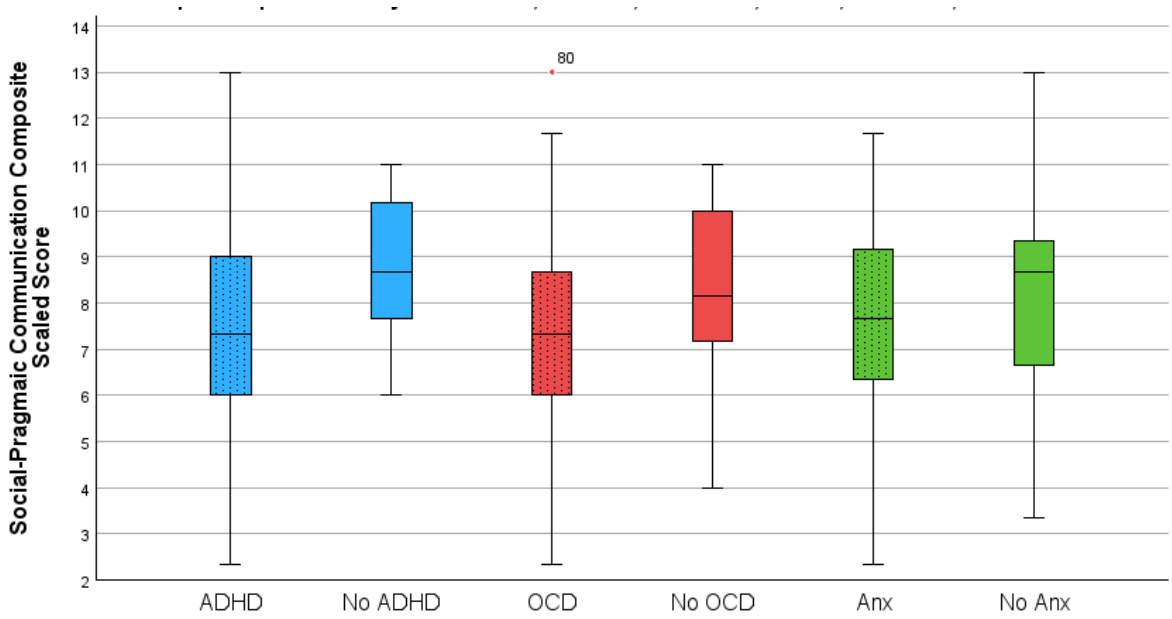


Figure 5

Box and Whisker Plots for Social-Pragmatic Communication for Children With and Without ADHD, OCD and Anxiety



Relationship Between Psychosocial Functioning and Communication Skills (Research Question 3)

A total of 29 participants had psychosocial difficulties in the 'high' or 'very high' range (48%) and 32 in the 'average" or 'slightly raised' range (52%). Among the 23 children with General Communication and/or Social-Pragmatic Communication scores more than 1 SD below the mean, 17 (74%) had high or very high psychosocial difficulties and six (26%) had average or slightly raised psychosocial difficulties. Among the 38 children with no communication concerns 12 (32%) had high or very high psychosocial difficulties, and 26 (68%) had average or slightly raised psychosocial difficulties. Table 6 shows the numbers and percentages of children with and without psychosocial difficulties and with and without communication difficulties by sex, age, and parent education. There were significant negative correlations between both composite

scores and the SDQ total difficulties score (psychosocial skills). General Communication: r = -.49, N = 61, p < .001, one tailed and Social-Pragmatic Communication: r = -.44, N = 61, p < .001, one tailed. So, as communication skills go up, the number of psychosocial difficulties go down and vice versa. These are medium-sized correlations. The scattergrams (Figures 6 and 7) show that the data points represent a linear relationship; distributed along the regression line with no outliers. Scattergrams of the relationship between psychosocial skills and communication skills for each tic disorder severity: low, medium, and high can be seen in Figures 8 and 9. Children with high tic disorder severities cluster in the top left of the scattergram with a combination of low communication skills and high psychosocial difficulties whereas children with low tic disorder severity cluster in the bottom right of the scattergram with a combination of high communication skills and low psychosocial difficulties.

Table 6

Sex, Age, and Parent Education Across Groups

	Sex		Age (mean)	Parent education	
	Male *n* (%) *n* = 41	Female *n* (%) *n* = 20		Completed high school *n* (%) *n = 5*	College/ university *n* (%) *n = 56*
Psychosocial difficulties					
High or very high *n* = 29	20 (49)	9 (45)	13.0	2 (40)	27 (48)
Average or slight *n* = 32	21 (51)	11 (55)	13.2	3 (60)	29 (52)

	Sex		Age (mean)	Parent education	
	Male *n* (%) *n* = 41	Female *n* (%) *n* = 20		Completed high school *n* (%) *n* = 5	College/ university *n* (%) *n* = 56
General Communication Composite					
<1SD *n* = 19	12 (29)	7 (35)	13.1	1 (20)	18 (32)
>/=1SD *n* = 42	29 (71)	13 (65)	13.0	4 (80)	38 (68)
Social-Pragmatic Communication Composite					
<1SD *n* = 21	14 (34)	7 (35)	13.2	2 (40)	19 (34)
>/=1SD *n* = 40	27 (66)	13 (65)	13.0	3 (60)	37 (66)

Figure 6

Scattergram of SDQ Total Difficulties Score by General Communication Composite Standard Score

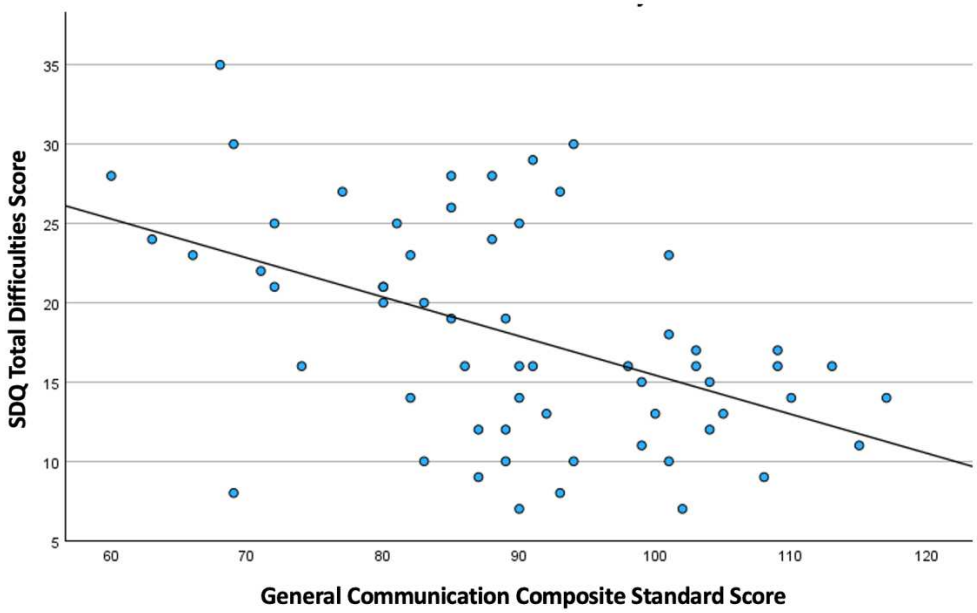


Figure 7

Scattergram of SDQ Total Difficulties Score by Social-Pragmatic Communication Composite Standard Score

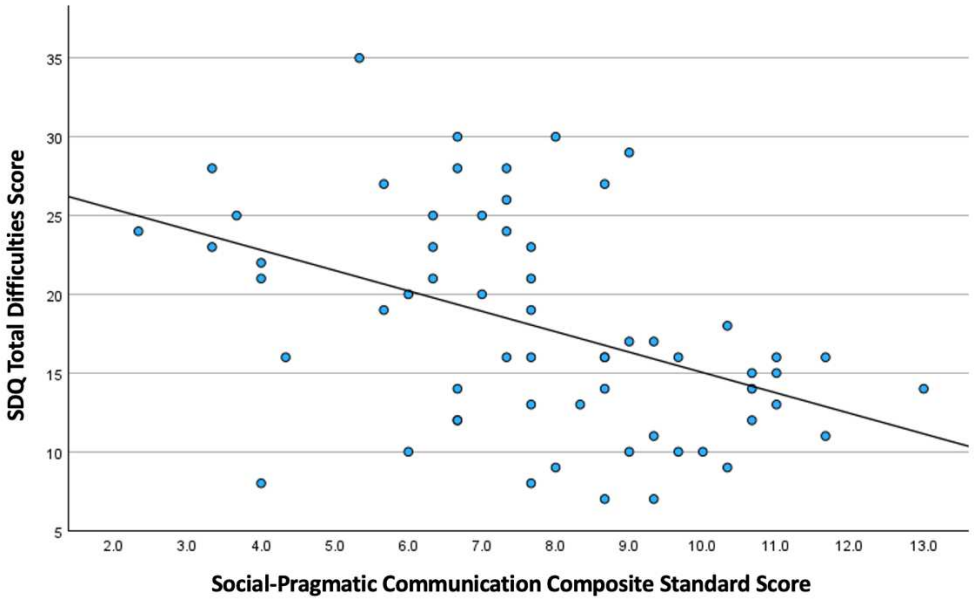


Figure 8

Scattergram of SDQ Total Difficulties Score by General Communication Composite Standard Score for Low, Medium, and High Tic Disorder Severity

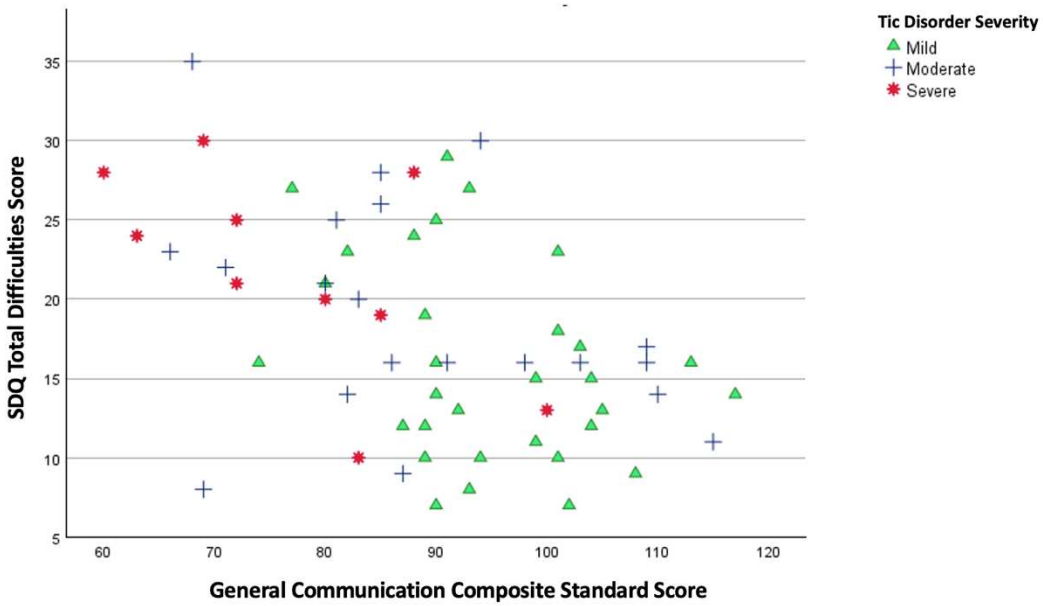
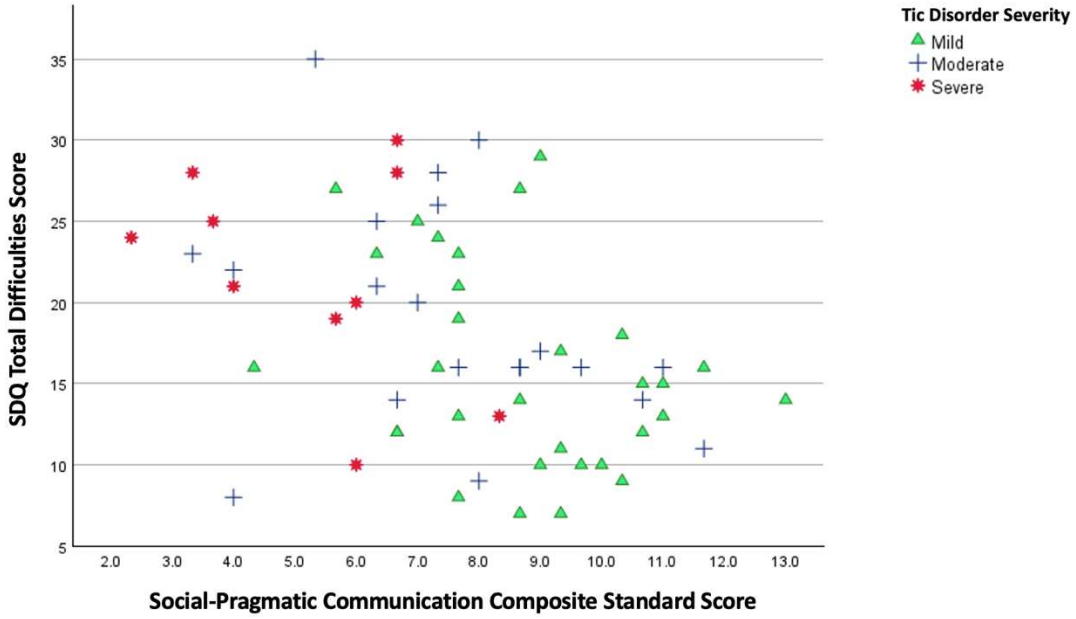


Figure 9

Scattergram of SDQ Total Difficulties Score by Social-Pragmatic Communication Composite Standard Score for Low, Medium, and High Tic Disorder Severity



Discussion

The purpose of this study was to describe the proportion of children in a TS sample who present with challenges in language and social communication on a parent questionnaire, whether these skills differ from normative samples, whether the presence of co-occurring conditions relates to these skills, and whether these skills are related to psychosocial functioning.

Communication Skills

We used a cut score of one SD below the mean (85) on the General Communication composite for identifying communication challenges in our sample. This cut score is expected to provide 83% positive predictive value for identifying both Developmental Language Disorder (termed Specific Language Impairment at the time) and Pragmatic Language Impairment (Bishop, 2003). Assuming this positive predictive power, we would expect that 16 of the 19

children with scores more than 1 SD below the mean have language disorders; however, only 6 had been diagnosed. This low identification rate is concerning given that the children in our sample were well into their school-aged years and therefore have presumably missed out on potential early intervention. This also raises questions about how, for some children, unidentified and unaddressed language/communication challenges could be contributing to the broad range of family, academic, and social problems that are reported in children with TS (Ricketts et al, 2022). In past research with complex TS samples, around 20% of children had been identified with language disorders (Cravedi et al., 2018; Spencer et al., 1998); however, children were not assessed directly to confirm language disorder rates.

Children with TS are diverse, and most children in our sample demonstrated average communication skills. General Communication Composite (GCC) means/medians in our sample were similar to children with emotional-behavioural disorders (median GCC = 88; Mackie & Law, 2010). Lower means have been documented in children with other conditions, like mood and anxiety disorders (GCC = 82), hyperactive symptoms (GCC = 78), inattentive symptoms (GCC = 66), Williams syndrome (GCC = 63), and Autism (GCC = 59) (Bignell & Cain, 2007; Philofsky et al., 2007; Towbin et al., 2005). When considering the means in our sample, it's important to note the wide range of scores that children received (domain Scaled Scores ranged from 1-16, $M = 10$, $SD = 3$). We cannot assume that all children with TS have lower communication skills than their peers; however, it is important to ensure that children experiencing difficulties are referred to a speech-language pathologist (SLP) for assessment and to receive appropriate intervention and accommodations to support communication development.

CCC-2 Domain Scores

Speech and Syntax. Scores in Speech and Syntax indicate that these children, as a whole, were not experiencing challenges with speech clarity or grammar. These results are not surprising given that previous research has demonstrated strengths in grammatical processing (Walensky, 2007).

Semantics. We expected that semantics would be an area of strength based on previous literature, however, the CCC-2 Semantics questions focus not just on word knowledge, but also on word use (including word-finding) in day-to-day communication contexts. Word-finding was reported as an area of challenge in case studies conducted several decades ago in the neuropsychology context (O'Quinn & Thompson, 1980; Thompson et al., 1979) and could be an area to focus on in future research using tools that focus specifically on word-finding, as the CCC-2 was not designed for this purpose.

Coherence. Coherence and Scripted Language were also significantly below expected norms for our sample as a whole. Coherence was one area of language challenges identified in early neuropsychological case studies (O'Quinn & Thompson, 1980; Thompson et al., 1979). Further, previously identified challenges with fluent expressive language formulation (de Nil et al., 2005; Donaher, 2008) would presumably affect the coherence of messages in communication.

Scripted Language. Lower scores in Scripted Language could potentially be the result of verbal tics during interactions and direct observations of children's communication would be needed in order to determine if these communicative features go beyond just tics.

Social-Pragmatic Communication: Initiation, Context and Nonverbal Communication. The three domains that comprised the Social-Pragmatic Communication

Composite all emerged as areas of challenge in our sample. It is important to consider how the onset of vocal and motor tics during interactions could affect initiation and could interrupt some elements of coordinated nonverbal communication, such as natural movements of the eyes, face, and arms as the person with TS is communicating. Similarly, tics could distract people with TS from the cues of their social partner, leading them to miss out on information being communicated. Direct observations of communication are needed in order to understand where differences in initiation and nonverbal communication lie and whether they go beyond tic-related differences in behaviour. Lower scores on questions in the Context domain could be explained by challenges in interpreting high-level language (e.g., inferences and non-literal language), as was reported by Legg et al. (2005). High-level language skills as well as other aspects of contextual communication are tied in with social cognition and several studies have concluded that individuals with TS exhibit social cognitive differences (e.g., Eddy et al., 2010a, 2010b, 2011, 2016, 2017; Eddy & Cavanna, 2015). Eddy has theorized that people with TS have to move through an additional layer of complexity when they interpret the social behaviour of others (Eddy 2021). Individuals with TS produce both voluntary and involuntary communicative information as they interact with others, but they must interpret the intentions of others who produce only voluntary communicative acts (although contrary information may be shared deliberately, as in sarcasm). This can lead to a greater number of misinterpretations and comprehension errors; however, it may only lead to a noticeable breakdown in the message when the information is more complex (i.e., high-level).

Social Relations and Interests. The Social Relations and Interests domains of the CCC-2 are thought to be associated with autistic characteristics (Saul et al., 2022). These were both areas of challenge in our sample. It is important to consider the ways in which living with tics

can complicate how one relates socially to others. Individuals with TS may make vocal noises, involuntarily move muscle groups used for verbal and nonverbal communication, use words that are involuntary, use socially inappropriate speech and gestures, and curse unintentionally during interactions. Since the interaction between social relations and experiences of social isolation and societal/self-stigma are complex to tease apart (Robertson & Eapen, 2017; Suh et al., 2022), treatments that reduce the symptoms of TS could lead to practical improvements in social relations for this group. Evidence-based interventions for tic disorders include psychoeducation, cognitive behavioural therapy, and medication (Martino & Leckman, 2022). There has also been much interest in improving the social experiences of children with TS by reducing stigma through educating peers, parents, teachers and the child with TS about TS and TS acceptance (Chowdhury & Christie, 2002; Fletcher et al., 2021; Ludlow et al., 2022; Mingbunjerdsuk & Zinner, 2020; Nussey et al., 2014; Wu & McGuire, 2018). Lower scores in the Interests domain for our sample indicate that children with TS may have a propensity toward preferring unique/specific activities and conversation topics, rote communication style, and predictability. These are characteristics commonly seen in autism but have also been described in other neurodivergent groups such as people with ADHD who are highly motivated by their specific topics of interest (Climie & Mastoras, 2015) and people with OCD who may prefer predictability (Williams et al., 2014). Whether these characteristics pose practical problems in communication or are just related to widely accepted neuronormative expectations in communication will be important to consider going forward.

Co-occurring Conditions

ADHD, OCD and anxiety are all conditions that are associated with communication challenges but past research looking at the contribution of specific conditions to these challenges

has produced inconsistent findings. While a few studies have found that ADHD and/or OCD co-occurrence may relate to language challenges (De Groot et al., 1997; Sukhodolsky et al., 2003), findings on social language have been inconsistent (Carter et al., 2000; Darrow et al; 2017; Pringsheim & Hammer, 2013; Sukhodolsky et al., 2003). We expected that the presence of ADHD, OCD and anxiety would contribute significantly to communication challenges; however, in this sample, this was not the case. Here, it appears that no one additional diagnosis on its own and no combination of these diagnoses increased the child's likelihood of having communication challenges. Our sample was composed almost entirely of children with multiple co-occurring conditions (nearly 90%), and the results are reflective of that. Past research has found that children with TS do not have elevated rates of language disorders when there are no co-occurring conditions (Cravedi et al., 2018; Spencer et al., 1998).

Psychosocial Skills in Relation to Communication

We saw that when language and social communication skills were lower, psychosocial challenges were higher and vice versa. We can also see that children with high tic disorder severities cluster toward the corner of the scattergram where low communication skills and high psychosocial challenges sit, and children with low tic disorder severity cluster toward the opposite corner (without communication or psychosocial challenges; Figures 8 and 9). The relationship between psychosocial difficulties and communication has not previously been examined in children with TS and this study has provided data to suggest a relationship between the two, but no conclusions regarding causation can be made. It is important to consider; however, that supporting language and communication development in this group of children through appropriate interventions could potentially contribute to positive psychosocial outcomes as well.

Limitations

The interpretation of results in this study is limited by the use of parent reports as the sole method of gathering information. In particular, more detailed and accurate information about co-occurring conditions could be obtained if the data had been collected through a healthcare provider who collects this medical information. The CCC-2 has been used as a measure of communication in research with a wide variety of clinical groups; however, some questions on the assessment may identify tics rather than communication difficulties. This possibility limits the interpretation of results and should be considered going forward when using this particular tool in research and clinical practice. Further, the use of convenience sampling means that our sample may not be representative of the population as a whole. One notable difference is in the number of children who had co-occurring conditions. Our sample had high levels of co-occurring conditions, with 98% reporting at least one condition and 89% reporting two or more conditions. Previous literature suggests that about 80% of children with TS have co-occurring conditions and 50% have two or more conditions (Cath et al., 2022).

Another limitation was the lack of representativeness when it came to family characteristics. The education level of respondents was high with over 90% having received a college/university education. This does not represent North American families as a whole, where approximately 69% of Canadians and 63% of Americans have a postsecondary degree or diploma (Statistics Canada, 2016; United States Census Bureau, 2022a). Similarly, the rate of multilingualism in our sample was low compared to Canadian (18%) and American (20%) rates (Statistics Canada, 2023a; United States Census Bureau, 2022b). A large proportion of families in our sample were White, making the number of racialized families low in comparison to

Canadian (27%) and American (24%) rates (Statistics Canada, 2023b; United States Census Bureau, 2023).

Future Research Directions

Our results indicate that language and communication challenges potentially fall across a range of domains (all domains of the CCC-2 except Speech and Syntax). For a deeper understanding of language and communication differences, future research should aim to investigate the language skills of children with TS directly rather than relying on parent reports. Understanding the lived experience of individuals with Tourette syndrome is of importance and therefore we must ask children about their communication and social interaction experiences. Also, direct observation of children's skills would be beneficial and could investigate word-finding skills, coherence in language construction, and aspects of contextual communication. In keeping with the important theme of TS acceptance, it will be important going forward to consider which aspects of communication in TS require our focus and intervention to improve quality of life and communicative function and which aspects require our acceptance and understanding. More specifically, future discussion is needed as to where language and communication differences in TS are functionally limiting for individuals with TS and where they may be identifiable only because they are inconsistent with neuronormative expectations. It is important to critically evaluate language research and intervention practices that may increase stigma or create further social barriers for people with TS.

Future research should also investigate the relationship between psychosocial skills and language/communication development further to better understand how these two variables are related. It will be important to understand whether supporting language and communication development in this population has positive effects on levels of overall psychosocial functioning.

It will also be important to investigate the relationship between communication development and presence of co-occurring conditions, tic disorder severity, and psychosocial skills beyond parent-reported methods.

Conclusion

This study has provided information about rates of communication concerns in a sample of children with TS. It has identified specific areas of development that may commonly present concerns in this population—particularly for children with co-occurring conditions. SLPs can use this information to guide assessment and as a starting point when thinking about intervention goals, although there is still much work to be done to clarify how to best support children with TS.

References

Albin, R. (2018). Tourette syndrome: A disorder of the social decision making network. *Brain, 141*(2), 332–347.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). https://doi.org/10.1176/appi.books.9780890425596

Bishop, D. (1989). *Children's Communication Checklist (CCC)*. Pearson.

Bishop, D. (2003). *Children's Communication Checklist (CCC-2)* (2nd ed.). Pearson.

Bignell, S., & Cain, K. (2007). Pragmatic aspects of communication and language comprehension in groups of children differentiated by teacher ratings of inattention and hyperactivity. *British Journal of Developmental Psychology*, *25*(4), 499–512. https://doi.org/10.1348/026151006X171343

Carter, A. S., O'Donnell, D. A., Scahill, L., Schultz, R. T., Leckman, J. F. & Pauls, D. L. (2000). Social and emotional adjustment in children affected with Gilles de la Tourette's Syndrome: Associations with ADHD and family functioning. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 41*(2), 215–223. https://doi.org/10.1017/S0021963099005156

Cath, D. C., Muller-Vahl, K. R., & Mathews, C. A. (2022). Other psychiatric comorbidities in Tourette syndrome. In D. Martino & J. F. Leckman (Eds.), *Tourette syndrome* (2nd ed., pp. 65–94). Oxford University Press.

Chowdhury, U., & Christie, D. (2002). Focus on practice: Tourette syndrome: A training day for teachers. *British Journal of Special Education*, *29*(3), 123–126. https://doi.org/10.1111/1467-8527.00254

Claussen, A. H., Bitsko, R. H., Holbrook, J. R., Bloomfield, J., & Giordano, K. (2018). Impact of Tourette syndrome on school measures in a nationally representative sample. *Journal of Developmental and Behavioral Pediatrics, 39*(4), 335–342. https://doi.org/10.1097/DBP.0000000000000550

Climie, E. A., & Mastoras, S. M. (2015). ADHD in schools: Adopting a strengths-based perspective. *Canadian Psychology, 56*(3), 295–300. https://doi.org/10.1037/cap0000030

Constantino, J. N., & Gruber, C. P. (2012). *Social Responsiveness Scale (SRS)* (2nd ed.). Western Psychological Services.

Cravedi, E., Deniau, E., Giannitelli, M., Pellerin, H., Czernecki, V., Priou, T., Xavier, J., Consoli, A., Hartmann, A. & Cohen, D. (2018). Disentangling Tourette syndrome heterogeneity through hierarchical ascendant clustering. *Developmental Medicine and Child Neurology, 60*(9), 942–950. https://doi.org/10.1111/dmcn.13 913

Darrow, S. M., Grados, M., Sandor, P., Hirschtritt, M. E., Illmann, C., Osiecki, L., Dion, Y., King, R., Pauls, D., Budman, C. L., Cath, D. C., Greenberg, E., Lyon, G. J., McMahon, W. M., Lee, P. C., Delucchi, K. L., Scharf, J. M. & Mathews, C. A. (2017). Autism spectrum symptoms in a Tourette's disorder sample. *Journal of the American Academy of Child and Adolescent Psychiatry, 56*(7), 610–617. https://doi.org/10.1016/j.jaac.2017.05.002

De Groot, C. M., Yeates, K. O., Baker, G. B., & Bornstein, R. A. (1997). Impaired neuropsychological functioning in Tourette's syndrome subjects with co-occurring obsessive-compulsive and attention deficit symptoms. *Journal of Neuropsychiatry and Clinical Neurosciences, 9*(2), 267–272. https://doi.org/10.1176/jnp.9.2.267

De Nil, L. F., Sasisekaran, J, Van Lieshout, P. H. H. M., & Sandor, P. (2005). Speech disfluencies in individuals with Tourette syndrome. *Journal of Psychosomatic Research, 58*(1), 97–102. https://doi.org/10.1016/j.jpsychores.2004.06.002

Donaher, J. (2008). *Speech fluency demonstrated by children with Tourette syndrome* (3320012) [Doctoral Dissertation, Temple University]. https://doi.org/10.1016/b978-012397720-5.50034-7

Eapen, V., McPherson, S., Karlov, L., Nicholls, L., Črnčec, R., & Mulligan, A. (2019). Social communication deficits and restricted repetitive behavior symptoms in Tourette syndrome. *Neuropsychiatric Disease and Treatment, 15*, 2151–2160. https://doi.org/10.2147/ ndt.s210227

Eddy, C. M. (2021) Tourette syndrome. In: Commings, L. (Ed.), *Handbook of pragmatic language disorders* (pp. 375–383). Springer.

Eddy, C. M., & Cavanna, A. E. (2013). Altered social cognition in Tourette syndrome: Nature and implications. *Behavioural Neurology, 27*(1), 15–22.

Eddy, C.M. & Cavanna, A.E. (2015) Triangles, tricks and tics: hypermentalizing in response to animated shapes in Tourette syndrome. *Cortex, 71*, 68–75.

Eddy, C. M., Cavanna, A. E. & Hansen, P. C. (2017). Empathy and aversion: The neural signature of mentalizing in Tourette syndrome. *Psychological Medicine, 47*(3), 507–517.

Eddy, C. M., Mitchell, I. J., Beck, S. R., Cavanna, A. E. & Rickards, H. E. (2010a). Altered attribution of intention in Tourette's syndrome. *Journal of Neuropsychiatry and Clinical Neurosciences, 22*(3), 348–351. https://doi.org/10.1176/jnp.2010.22.3.348

Eddy, C. M., Mitchell, I. J., Beck, S. R., Cavanna, A. E. & Rickards, H. E. (2010b). Impaired comprehension of nonliteral language in Tourette syndrome. *Cognitive and Behavioral Neurology, 23*(3), 178–184.

Eddy, C. M., Mitchell, I. J., Beck, S. R., Cavanna, A. E. & Rickards, H. (2011). Social reasoning in Tourette syndrome. *Cognitive Neuropsychiatry, 16*(4), 326–47.

Eddy, C. M., Cavanna, A. E., Rickards, H. E. & Hansen, P. C. (2016). Temporo-parietal dysfunction in Tourette syndrome: Insights from an fMRI study of theory of mind. *Journal of Psychiatric Research, 81*, 102–11.

Ehlers, S., Gillberg, C., Wing, L. (1999). A screening questionnaire for Asperger syndrome and other high-functioning autism spectrum disorders in school age children. *Journal of Autism and Developmental Disorders, 29*, 12–141.

Feehan, A., & Charest, M. (2023). A scoping review of oral language and social communication abilities in children with Tourette syndrome. *International Journal of Language and Communication Disorders, 59*(1), 1–22. https://doi.org/10.1111/1460-6984.12949

Fisher, A. P., Gies, L. M., Chapman, L., Aguilar, J. M., Yeates, K. O., Taylor, H. G., & Wade, S. L. (2022). The clinical utility of the Children's Communication Checklist-2 in children with early childhood traumatic brain injury. *Clinical Neuropsychologist, 36*(7), 1728–1745. https://doi.org/10.1080/13854046.2020.1866675

Fletcher, J., Dimitropoulos, G., Martino, D., Wilcox, G., MacMaster, F., Arnold, P., & Pringsheim, T. (2021). Developing a provincial patient support network for children and families affected by Tourette syndrome and/or obsessive–compulsive disorder: Results of a stakeholder consultation. *Child and Adolescent Psychiatry and Mental Health, 15*(1), 1–10. https://doi.org/10.1186/s13034-021-00383-5

Gadow, K.D. & Paolicelli, L. M. (1986). *The Global Tic Rating Scale (GTRS)*. State University of New York Department of Psychiatry.

Ghahari, S., Hassani, H., & Purmofrad, M. (2017). Pragmatic competency and Obsessive–Compulsive Disorder: A comparative assessment with normal controls. *Journal of Psycholinguistic Research, 46*(4), 863–875. https://doi.org/10.1007/s10936-016-9467-6

Goodman, R. (1997). *Strengths and Difficulties Questionnaire (SDQ)* [Database record].

Güler, A.S., Berkem, M., Yazgan, Y., & Kalaça, S. (2015). Cognitive flexibility and social responsiveness in children and adolescents with Tourette syndrome. *Child Psychiatry and Human Development, 46*(6), 940–950. https://doi.org/10.1007/s10578-015-0533-3

Gutierrez-Colina, A. M., LaMotte, J., Eaton, C., Kardon, P., & Blount, R. L. (2015). The role of self-competence in health-related quality of life and behavioral functioning of children with Tourette syndrome. *Journal of Developmental and Behavioral Pediatrics, 36*(9), 743–751. https://doi.org/10.1097/DBP.0000000000000214

Helland, W. A., Helland, T., & Heimann, M. (2014) Language profiles and mental health problems in children with specific language impairment and children with ADHD. *Journal of Attention Disorders, 18*(3), 226-235. https://doi.org/10.1177/1087054712441705

Hulbert, T. (1986) Neuropsychological subgrouping of Tourette children. [Unpublished doctoral dissertation]. Wayne State University. https://doi.org/10.1017/CBO9781107415324.004

Kadesjö, B. & Gillberg, C. (2000). Tourette's disorder: Epidemiology and comorbidity in primary school children. *Journal of the American Academy of Child and Adolescent Psychiatry, 39*(5), 548–555. https://doi.org/10.1097/00004583-200005000-00007

Korrel, H., Mueller, K. L., Silk, T., Anderson, V., & Sciberras, E. (2017). Research review: Language problems in children with Attention-Deficit Hyperactivity Disorder: A systematic meta-analytic review. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 58*(6), 640–654. https://doi.org/10.1111/jcpp.12688

Legg, C., Penn, C., Temlett, J., & Sonnenberg, B. (2005). Language skills of adolescents with Tourette Syndrome. *Clinical Linguistics and Phonetics, 19*(1), 15–33. https://doi.org/10.1080/ 02699200410001665270

Ludlow, A. K., Cutler, A., & Keville, S. (2022). British teachers' knowledge and awareness of Tourette syndrome in children attending mainstream school. *Journal of Research in Special Educational Needs, 22*(4), 323–331. https://doi.org/10.1111/1471-3802.12570

MacKie, L., & Law, J. (2010). Pragmatic language and the child with emotional/behavioural difficulties (EBD): A pilot study exploring the interaction between behaviour and communication disability. *International Journal of Language and Communication Disorders, 45*(4), 397–410. https://doi.org/10.3109/13682820903105137

Martino, D., & Leckman, J. F. (2013). *Tourette syndrome*. Oxford University Press.

Martino, D., & Leckman, J. F. (2022). *Tourette syndrome* (2nd ed.). Oxford University Press.

Martino, D., Pringsheim, T. M., Cavanna, A. E., Colosimo, C., Hartmann, A., Leckman, J. F., Luo, S., Munchau, A., Goetz, C. G., Stebbins, G. T., Martinez-Martin, P. (2017). Systematic review of severity scales and screening instruments for tics: Critique and recommendations. *Movement Disorders, 32*(3), 467–473. https://doi.org/10.1002/mds.26891

Mingbunjerdsuk, D., & Zinner, S. H. (2020). Coping with Tics in School and with Peers. *Current Developmental Disorders Reports, 7*(4), 227–236. https://doi.org/10.1007/s40474-020-00218-6

Norbury, C., Gooch, D., Wray, C., Baird, G., Charman, T., Simonoff, E., Vamvakas, G., & Pickles, A. (2016). The impact of nonverbal ability on prevalence and clinical presentation of language disorder: Evidence from a population study. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 57*(11), 1247-1257.

Nussey, C., Pistrang, N., & Murphy, T. (2014). Does it help to talk about tics? An evaluation of a classroom presentation about Tourette syndrome. *Child and Adolescent Mental Health, 19*(1), 31–38. https://doi.org/10.1111/camh.12000

O'Quinn, A. N. & Thompson, R. J. (1980). Tourette's syndrome: An expanded view. *Pediatrics, 66*(3), 420–424. https://doi.org/10.1016/ s0002-7138(09)61008-6

Philofsky, A., Fidler, D. J., & Hepburn, S. (2007). Pragmatic language profiles of school-age children with autism spectrum disorders and Williams Syndrome. *American Journal of Speech-Language Pathology, 16*(4), 368–380. https://doi.org/10.1044/1058-0360(2007/040)

Pringsheim, T., & Hammer, T. (2013). Social behavior and comorbidity in children with tics. Pediatric Neurology, 49(6), 406–410. https://doi.org/10.1016/j.pediatrneurol.2013.08.005

Redmond, S. M. (2004). Conversational profiles of children with ADHD, SLI and typical development. *Clinical Linguistics and Phonetics, 18*(2), 107–125. https://doi.org/10.1080/02699200310001611612

Robertson, M. M., & Eapen, V. (2017). The psychosocial aspects of the Gilles de la Tourette syndrome: Empirical evidence from the literature. *Current Behavioral Neuroscience Reports, 4*(1), 59–69. https://doi.org/10.1007/s40473-017-0107-8

Rutter, M., Bailey, A., & Lord, C. (2003). *The Social Communication Questionnaire (SCQ).* Western Psychological Services.

Saul, J., Griffiths, S., & Norbury, C. F. (2022). Prevalence and functional impact of social (pragmatic) communication disorders. *Journal of Child Psychology and Psychiatry and Allied Disciplines,* 1–12. https://doi.org/10.1111/jcpp.13705

Spencer, T., Biederman, J., Harding, M., O'Donnell, D., Wilens, T., Faraone, S., Coffey, B., & Geller, D. (1998). Disentangling the overlap between Tourette's disorder and ADHD. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 39*(7), 1037–1044. https://doi.org/10.1017/S0021963098002984

Statistics Canada (2023b). *Changing demographics of racialized people in Canada*. https://www150.statcan.gc.ca/n1/pub/36-28-0001/2023008/article/00001-eng.htm

Statistics Canada (2023a). *English–French bilingualism in Canada: Recent trends after five decades of official bilingualism*. https://www12.statcan.gc.ca/census-recensement/2021/as-sa/98-200-X/2021013/98-200-X2021013-eng.cfm

Statistics Canada (2016). *More post-secondary education indicators*. https://www150.statcan.gc.ca/n1/en/subjects/education_training_and_learning/postsecondary_education

Suh, H. W., Yoon, S. I., Hong, S., Lee, H. W., Lee, M., Kim, J. W., & Chung, S. Y. (2022). How do children with Tourette's syndrome and their caregivers live with the disorder? A

systematic review of qualitative evidence. *Frontiers in Psychiatry, 13*(September), 1–16. https://doi.org/10.3389/fpsyt.2022.992905

Sukhodolsky, D. G., Scahill, L., Zhang, H., Peterson, B. S., King, R. A., Lombroso, P. J., & Thompson, R. J., O'Quinn, A. N., & Logue, P. E. (1979). Gilles de la Tourette syndrome: A review and neuropsychological aspects of four cases. *Journal of Pediatric Psychology, 74*(4), 371–387. https:// doi.org/10.1177/014107688607900508

Towbin, K. E., Pradella, A., Gorrindo, T., Pine, D. S., & Leibenluft, E. (2005). Autism spectrum traits in children with mood and anxiety disorders. *Journal of Child and Adolescent Psychopharmacology, 15*(3), 452–464. https://doi.org/10.1089/cap.2005.15.452

United States Census Bureau (2022a). *Census Bureau releases new educational attainment data.* https://www.census.gov/newsroom/press-releases/2022/educational-attainment.html

United States Census Bureau (2022b). *What languages do we speak in the United States.* https://www.census.gov/library/stories/2022/12/languages-we-speak-in-united-states.html

United States Census Bureau (2023) *Quick facts*. https://www.census.gov/quickfacts/fact/table/US/PST045223

Vaïsänen, R., Loukusa, S., Moilanen, I., & Yliherva, A. (2014). Language and pragmatic profile in children with ADHD measured by children's communication checklist 2nd edition. *Logopedics Phoniatrics Vocology, 39*(4), 179–187. https://doi.org/10.3109/14015439.2013.784802

Van Steensel, F. J. A., Bögels, S. M., & Wood, J. J. (2013). Autism spectrum traits in children with anxiety disorders. *Journal of Autism and Developmental Disorders, 43*(2), 361–370. https://doi.org/10.1007/s10803-012-1575-z

Verté, S., Geurts, H. M., Roeyers, H., Oosterlaan, J., & Sergeant, J. A. (2005). Executive functioning in children with autism and Tourette syndrome. *Development and Psychopathology, 17*(2), 415–445. https://doi.org/10.1017/S0954579405050200

Wadman, R., Glazebrook, C., Beer, C. & Jackson, G. M. (2016). Difficulties experienced by young people with Tourette syndrome in secondary school: A mixed methods description of self, parent and staff perspectives. *BMC Psychiatry, 16*(1), 1–11. https://doi.org/10.1186/s12888-016-0717-9

Walenski, M., Mostofsky, S. H. & Ullman, M. T. (2007). Speeded processing of grammar and tool knowledge in Tourette's syndrome. *Neuropsychologia, 45*(11), 2447–2460. https://doi.org/10. 1016/j.neuropsychologia.2007.04.001

Williams, M. T., Mugno, B., Franklin, M., & Faber, S. (2014). Symptom dimensions in Obsessive-Compulsive Disorder: Phenomenology and treatment outcomes with exposure and ritual prevention. *Psychopathology, 46*(6), 365–376. https://doi.org/10.1159/000348582.

Wu, M. S., & McGuire, J. F. (2018). Psychoeducation about tic disorders and treatment. In J. F. McGuire, T. K. Murphy, J. Piacentini, & E. A. Storch (Eds.), *The clinician's guide to treatment and management of youth with Tourette syndrome and tic disorder* (pp. 21–41). Elsevier Academic Press. https://doi.org/10.1016/B978-0-12-811980-8.00002-9

Chapter 4: Methods Supplement for Communication and Psychosocial Functioning in Children with Tourette Syndrome: Parent-Reported Measures (Study 2)

This chapter provides supplementary information about Study 2, described in the previous chapter. It describes the selection of specific measures for the study in detail and provides detailed information about the psychometric properties of the measures.

Selecting Measures of Communication, Psychosocial Function, and Tic Severity

Communication

The Children's Communication Checklist, Second Edition (CCC-2; Bishop, 2003) was selected as a measure of communication because it could be completed by parents, provides normative data for the 8-16 year age range, and yields individual domain scores as well as covering aspects of core language development (i.e., Syntax and Semantics), expressive formulation (i.e., Coherence), high-level language (i.e., Context), social-pragmatic language (i.e., Initiation, Context, Nonverbal Communication), and autistic communication characteristics (i.e., Social Relations and Interests). Additionally, composite scores for general communication and social-pragmatic communication can be derived. The CCC-2 demonstrates adequate specificity (.85; Timler, 2014) and results correlate moderately with a gold standard assessment (The Clinical Evaluation of Language Fundamentals, Fourth Edition; Kelso et al., 2012). Eight of the ten subscales have been shown to differentiate children with ADHD (a similar clinical population to TS) from typically developing children (Helland et al., 2014). The limitations of this tool are that sensitivity levels have been variable (i.e., low levels of sensitivity are reported in the examiner manual for detecting Specific Language Impairment; high levels are reported by

Timler (2014) for detecting language impairment in children with ADHD), standard error of measurement values are not available, studies of content and construct validity are limited (but consistent with other tests in this field), and study of the psychometric properties for test subscales is currently limited. The comparability between the CCC-2 normative sample and the TS survey sample can be seen in Table 7. Notable differences between the two samples are that the survey sample was largely made up of Canadian respondents rather than American respondents, comprised a greater number of male children, and comprised a greater number of parents with four or more years of postsecondary education. Overall, The TS survey sample was also less diverse with regard to race/ethnicity. See additional information about the psychometric properties of the CCC-2 in Table 8.

Table 7

Comparison Between the Survey Sample and the CCC-2 Normative Sample

	TS Survey Sample *N* = 61 *n*(%)	CCC-2 Normative Sample *N* = 950 *n*(%)
Canadian	43(70)	0(0)
American	18(30)	950(100)
Gender		
Male	41(67)	475(50)
Female	20(33)	475(50)
Parent education		
Did not complete high school	0(0)	144(15)
High school degree or equivalent	5(8)	295(31)
One to three years of postsecondary	16(26)	277(29)
Four or more years of postsecondary	40(66)	234(25)
Race/Ethnicity		
White	51(84)	588(62)
Other	10(16)[a]	362(38)[b]
Primary language spoken in the home		
English	57(93)	950(100)

	TS Survey Sample *N* = 61 *n*(%)	CCC-2 Normative Sample *N* = 950 *n*(%)
Other	4(7)[c]	0(0)[d]
English-only homes	52(85)	770(81)
Multilingual homes	9(15)	180(19)
Diagnosed with a speech/language condition/receiving services	11(18)	95(10)

[a]Race/ethnicities included: Japanese, Jewish, Filipino, Black, Indigenous, Latin American, South Asian, and Southeast Asian.

[b]Race/ethnicities included: Hispanic, African American, and other.

[c]Languages included: Arabic, Bengali, French, Spanish, and Ukrainian.

[d]Languages included: Spanish, Asian languages, and other.

Table 8

Psychometric Properties of the CCC-2

Validity/Reliability Criteria[a]	Test information	Evaluation
Classification accuracy		
Sensitivity and specificity	A cut-off of -1 SD using the General Communication Composite yielded a sensitivity of .70 and a specificity of .85 for detecting specific language impairment (SLI; term used in manual). Timler (2014) - Examined the classification accuracy of children with and without language impairment in a group with ADHD. Sensitivity was 1.0, specificity was .85.	Sensitivity ranges from low to high and specificity has been demonstrated to be adequate.
Negative and positive likelihood ratios	Not reported	Unknown

Validity/Reliability Criteria[a]	Test information	Evaluation
Similarity of the normative sample to the research sample		
Report of normative sample characteristics	950 US children were used for the normative sample, 19% of the sample spoke an additional language at home and 27% of the sample were receiving special support for language or other needs. Half of the standardization sample were male and half were female. The composition of the sample varied in terms of geographic locations across the USA, parent education, and race/ethnicity (including primarily White, Hispanic, and African American children).	Used a large sample that includes a diverse range of geographic locations, race/ethnicities, abilities, and parent education levels. The sample includes both males and females and bilingual children. The norming sample differs from the study population because it represents the US, not Canada.
Consistency in test results		
Consistency in administration (reliability)	The examiner's manual provides detailed instructions about the administration of the questionnaire. The test was standardized with parents completing the checklist.	The information is adequate so that the examiner can administer the test in the same way it was administered with the normative sample.
Test-retest reliability	The results of the test-retest reliability study found coefficients that range from .86-.96 (Pearson product-moment correlation coefficient).	Coefficients demonstrated good test-retest reliability.
Interrater reliability	Interrater reliability was assessed on an older version of the test; only items with high interrater reliability were carried forward.	The previous version indicates that interrater reliability is good.

Validity/Reliability Criteria[a]	Test information	Evaluation
Internal consistency	Internal consistency was calculated. Coefficient alpha ranged from .69-.85 for the CCC-2 scales.	Alpha values are near or above .7, which is considered an acceptable level of internal consistency.
Standard error of measurement	Not reported.	Unknown how much a participant's true score could vary from the score that they have obtained on the test.
Test measures the skill of interest		
Content reviewed by experts	None reported. Content was developed by consulting literature on language and pragmatic language development.	Some evidence of content validity.
Appropriateness of response process	Parents respond to each item while considering their observations of their child.	The response format is appropriate.
Comparison to a gold standard (criterion validity)	None in the CCC-2 manual Kelso et al. (2012) - found a moderate correlation between scores on the CCC-2 and the CELF-4 (Semel et al., 2003).	Moderate correlation with a 'gold standard' measure.
Structural validity	No factor analysis completed.	Unknown.

[a]Criteria based on Daub et al., 2021

Psychosocial Function

The Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997) was selected to measure psychosocial function because it could be completed by parents and it provides a total score for the level of psychosocial function of children in the 8-16 years range. It has been used in a wide variety of children's health research including in pediatric audiology, Autism Spectrum Disorders, Attention Deficit/Hyperactivity Disorders, and type 1 diabetes (e.g., de Jong et al., 2023; Kjærandsen et al., 2023; Marks et al., 2023). The SDQ considers emotional symptoms, conduct problems, hyperactivity/inattention, and peer relationship problems. Sensitivity for predicting emotional-behavioural disorders varies across studies (.63-.98; Goodman et al., 2000; 2004). The SDQ has a good negative predictive value (93.6; Bourdon et al., 2005). The limitations of this tool are that specificity for detecting emotional-behavioural disorders is not known and the positive predictive value is poor (45.0; Bourdon et al., 2005). See additional information about the psychometric properties in Table 9.

Table 9

Psychometric Properties of the SDQ

Validity/reliability criteria[a]	Test information	Evaluation
Classification accuracy criteria		
Classification accuracy	Goodman et al. (2000) - Associations between diagnosis and SCQ predictions were .6. High difficulties group predicted service contact: PPV = 45.0; NPV = 93.6 (Bourdon et al., 2005). Bourdon et al. (2005) recommend three methods of identifying "serious difficulties" 1) Total difficulties scale is in the top 10%, 2) parent identified definite or severe difficulties for "overall difficulties" question, or 3) impairment score is high AND high scores on emotional symptoms, conduct problems or inattention-hyperactivity.	Classification accuracy is fair, negative predictive value is good[a]; positive predictive value is poor[a].
Sensitivity and specificity	Goodman et al. (2000) - Overall sensitivity for detecting any emotional-behavioural disorder was .63. Goodman et al. (2004) - sensitivities for predicting diagnoses ranged from .83 to .98.	Sensitivity ranges from low to high. Specificity is not known.
Negative and positive likelihood ratios	No studies.	Unknown.
Similarity of the normative sample to the research sample		
Report of normative sample characteristics	The normative sample represents a randomly selected subset of the US population. Completed by parents of 9,878 children.	The population is similar.

Validity/reliability criteria[a]	Test information	Evaluation
Consistency in test results		
Consistency in administration (reliability)	Detailed instructions for administration and scoring.	The measure is consistent.
Test-retest reliability	Parent's reports were stable over 12 weeks (Borg et al., 2012).	Test-retest reliability is good.
Interrater reliability	Spearman Rho for agreement between parents was .65 (Borg et al., 2012).	Interrater reliability is good.
Internal consistency	Chronbach's Alpha ranged from .77-.86 (Borg et al., 2012). Chronbach's Alpha for Difficulties .83; Impairment scores .80; subscales .46–.77 (Bourdon et al., 2005).	Internal consistency fair to excellent for subscales; excellent for total difficulties; excellent for impairment scores.
Standard error of measurement	None.	Not available.
Test measures the skill of interest		
Content reviewed by experts	Unknown.	Unknown.
Appropriateness of response process	Parents respond to each item while considering their observations of their child in the last 6 months.	The response format is appropriate.

Validity/reliability criteria[a]	Test information	Evaluation
Comparison to a gold standard (criterion validity)	Achenbach et al. (2008) - reviewed studies that have correlated SDQ scores with scores on the Child Behaviour Checklist. The mean correlation across subtests ranged from .67- .75 (Becker et al., 2004; Goodman & Scott, 1999; Klasen et al., 2000; Van Widenfelt et al., 2003).	Moderate correlations with gold standard measure.
Structural validity	Factor analysis supported the five SDQ subscales (Woerner et al., 2004).	Evidence of structural validity.

[a]Criteria is based on Daub et al., 2021.

Tic Disorder Severity

The Global Tic Rating Sale (GTRS; Gadow & Paolicelli, 1986) provides a short series of questions measuring the current severity of the child's tic disorder from the parent's perspective (over the past week). This tool was "suggested" in a systematic review of tic severity scales and instruments (Martino et al., 2017). The strengths of this scale are that it is brief and easy to administer. The psychometric properties of the tool have not been evaluated.

Piloting the Questionnaires

The REDCap version of the questionnaires was piloted in two phases. The first phase involved sending the survey to four professionals with research and/or clinical training in communication sciences to seek feedback and corrections. After the suggested changes were made, the second phase involved sending the survey to three parents of children within the 8-16 years range who had communication and/or neurodevelopmental concerns (other than TS) to seek feedback and corrections. The updated survey represents the final version sent to participants.

Chapter 5: Language and Communication in Children with Complex Tourette Syndrome: Insights from Five Mixed Methods Case Studies (Study 3)

Abstract

Background. Children with Complex Tourette syndrome (C-TS) may present with challenges in their communication development, yet there is little research to guide clinicians working with this population.

Methods. This scaffolded mixed methods study aimed to understand and describe language development and day-to-day communication in five children with C-TS. Data were collected using the Global Tic Rating Scale (GTRS), the Strengths and Difficulties Questionnaire (SDQ), the Clinical Evaluation of Language Fundamentals, Fifth Edition, (CELF-5), the Test of Narrative Language, Second Edition (TNL-2), the Comprehensive Assessment of Spoken Language, Second Edition (CASL-2), the Children's Communication Checklist, Second Edition (CCC-2), communication log books, and semi-structured qualitative interviews. Communication was considered using the bioecological model as a framework with a focus on 'microstructure' contexts (i.e., school, home, and peer socialization contexts).

Results. All children had accessed or were accessing school accommodations, extra services, or alternative education options. In some cases, parents describe how children's educational environments language development and day-to-day communication. Managing family interaction in the home required parents to be thoughtful and proactive in their planning. Parents strategized to keep the home environment calm and productive, adopted attitudes of

acceptance and understanding, and accessed outside tools and supports. In some cases, parents provided social coaching and created intentional social experiences so that their child could be successful; in other cases, children were thriving socially without much need for parental intervention.

Conclusions. The experiences of children with C-TS are diverse. Interventions and accommodations need to be individualized to optimize language development and day-to-day communication.

Introduction

Tourette syndrome (TS) is a neurodevelopmental disorder involving motor and vocal tics that begin in childhood (American Psychological Association, 2013). About 80% of people with TS have co-occurring conditions (Cath et al., 2022), or 'complex' cases of TS (Complex TS; C-TS). Common co-occurring conditions complicating TS include attention-deficit/ hyperactivity disorder (ADHD), obsessive-compulsive disorder (OCD), anxiety, and learning disorders. While research has shown that many children with C-TS develop strong language skills, around 20% may have language disorders (Cravedi et al., 2018; Spencer et al., 1998).

Literature Measuring Communication Skills

Literature measuring communication skills in children with C-TS indicates that possible areas of difficulty include verbal IQ, tasks involving semantics (i.e., categorization and naming), social interaction, and complex social cognitive tasks (i.e., recognizing when there is a mismatch between vocal tone and the message being communicated; Carter et al., 2000; de Groot et al., 1997; Drury et al., 2012; Schuerholz et al., 1998; Sukhodolsky et al., 2003). Most of these studies do not report results specific to children with C-TS, but combine children with C-TS together with children who have 'uncomplicated' or 'pure' TS. A recent scoping review identified that children with TS tend to have communication profiles with many areas of strength (Feehan & Charest, 2023). Specifically, they appear to do well with language tasks that require processing at the single-word level (e.g., verbal fluency and vocabulary tests; Channon et al., 2003; Jensen, 2004). There is some (mostly dated) research that suggests children with TS may experience difficulty with following directions, language formulation, narrative language, word-finding, and high-level language such as inferencing and understanding non-literal language (Hulbert, 1986; Legg et al., 2005; O'Quinn & Thompson, 1980; Thompson et al., 1979). Also,

high levels of autistic traits and challenges in social communication have been documented using the Social Communication Questionnaire (Rutter et al., 2003) and the Social Responsiveness Scale (Constantino & Gruber, 2012; Eapen et al., 2019; Güler et al., 2015). These results highlight the importance of looking beyond language fundamentals such as vocabulary to understand where communication needs lie, thereby investigating skills such as language coherence, social communication, narrative language, and high-level language (e.g., inferencing and non-literal language).

TS in School, Home, and Peer Interaction Contexts

Communication functioning in children with TS in the school context has not previously been specifically investigated. Literature about school functioning generally indicates that rates of school problems are lower in children with uncomplicated TS and higher for children with TS and ADHD. In a nationally representative US sample, half of children with TS and co-occurring ADHD experience problems with overall school performance, 40% have reading problems, and 60% have writing problems (Ricketts et al., 2022). A study conducted in the UK found that teachers in primary and secondary mainstream schools had a poor understanding of TS, its co-occurring conditions, and how to support children with TS in the classroom (Ludlow et al., 2022). There is virtually no research looking into learning in children with TS who are homeschooled; however, an internet search revealed that some parents and organizations endorse this learning environment for children when the child is having difficulties in the school environment (e.g., Fitcher, 2014; Tourette Association of America, 2021). Previous qualitative research exploring parent perspectives of their children's experiences at school emphasized the need for accommodations in the classroom, the importance of being able to leave the classroom when needed, and the need for more understanding of how tics interfere with learning (Pine et

al., 2022). Classroom presentations educating peers about TS have been shown to improve peer attitudes about the condition (Nussey et al., 2014).

The communication functioning of children with TS in the home context has not been specifically investigated in past research; however, a range of challenges in the home context have been documented for both children and parents, affecting relationships, daily activities, and overall well-being (Cavanna et al., 2013, Cooper et al., 2003). Stress for parents is exacerbated when co-occurring conditions are present (Robinson et al., 2013; Stewart et al., 2015) and parents tend to feel more supported when others validate their experiences, recognize their efforts, and acknowledge their child's identity beyond TS (Travis & Juarez-Paz, 2020).

A few studies have explored children's experiences with social interaction. Research investigating the experiences of adolescents with TS suggests that negative social responses to tics from teachers and peers can be very upsetting: Young people may withdraw socially, become socially isolated, and experience loneliness, guilt, and shame (Jeans, 2016; Wadman et al., 2016). Planning to avoid things that trigger tics and keeping a distance in certain social situations allows young people to maintain an image of being "normal," but "good" friends are accepting and provide supportive responses to tics (Lee et al., 2016, p. 467). Social isolation is a common stressor in children with TS and has been connected to societal misunderstandings of the disorder, prejudice/stigma, and many other complex factors (Suh et al., 2022). Approximately one-third of children with uncomplicated TS and two-thirds of children with TS and ADHD may have problems relating socially to others (i.e., parents/siblings/peers; Ricketts et al., 2022). The severity level of a child's TS and the presence of co-occurring ADHD may affect their interpersonal well-being (Ricketts et al., 2022).

In summary, research investigating communication development in children with C-TS, thus far, has focused on testing specific areas of skill (e.g., vocabulary), but has not focused on communication success in day-to-day contexts (i.e., the use of language skills and communication in real-life settings), such as in the home, at school, and with peers. Measurement of communication skills and consideration of success in day-to-day communication contexts are both important in developing a holistic understanding of communication (Hyter, 2017). Past research provides initial clues as to where language strengths and needs may lie for children with C-TS; however, there is virtually no information about how children's communication development relates to their day-to-day contexts: school, the home environment, and peer socialization. This study used a mixed methods approach to collect information related to communication development in cases of C-TS both *quantitatively* through norm-referenced language and communication measures and *qualitatively* by investigating day-to-day communication in school, home, and peer contexts from the perspectives of parents. For the five case studies, we explored how different contexts and factors interact with language development and communication functioning—both impeding and facilitating them. The purpose of this study was to provide information that SLPs can draw on to support children with C-TS on their caseloads and when working as part of a medical or educational team. To consider both of these aspects of communication and provide a more holistic perspective on communication among children with C-TS, we drew on the bioecological model (Bronfenbrenner, 1979), whereby the characteristics of the child (e.g., communication skills) are presumed to interact with different child's day-to-day life contexts (the child's 'micro' contexts, such as school, home, and peer interactions). The use of this model provided a framework for understanding how

each child with their unique cognitive and language profile interacted with various day-to-day contexts in which they developed language skills and communicated.

Methods

Methodology

We were guided by two interpretive frameworks in this research, pragmatism and social constructionism. Pragmatists focus on selecting research methods that will best answer the research question of interest even if those methods come from different research traditions (i.e., quantitative and qualitative traditions; Creswell & Plano Clark, 2018). The qualitative component of this study was guided by social constructionism, the perspective that knowledge and reality are subjective, changing, and multiple, rather than objective, static, and singular (Crotty, 1998; Lincoln & Guba, 1985). Knowledge and reality are assumed to be built on our social systems, including linguistic, cultural, and socio-political contexts. Informed by these frameworks, five mixed methods case studies were used to construct information about the day-to-day communication of children with C-TS. The project approached understanding communication in C-TS from a strengths-based and functional standpoint (Bates, 1991) in contrast to a deficit-oriented approach that focuses on individual impairments. Communication was considered holistically with a focus on day-to-day communication rather than relying heavily on objective measures of communication (i.e., language and communication scores). A focus was placed on the contexts of the 'micro-system' (i.e., school, home, peer relations) within the bioecological perspective (Bronfenbrenner, 1979), as these are bioecological contexts in which the child engages in day-to-day communication. This scaffolded mixed methods project (Fetters, 2020; 2022) drew on: (a) case study methodology (Stake, 2006; 1995), (b) Interpretive Description (Thorne, 2016) by incorporating the disciplinary perspective of speech-language

pathology into study design and conduct, and (c) naturalistic inquiry (Lincoln & Guba, 1985) with a focus on understanding communication in naturally occurring settings. This research was approved by the institutional Human Research Ethics Board, Pro00125196.

Participant Recruitment

Information about the study was shared via advertisements on social media and regional Tourette-focused organizations in a major Canadian city. Purposeful sampling was used to select five children with C-TS and one of their parents to participate. Children were included if they were 8-12 years of age, if they did not have intellectual disabilities or autism, and if they had past or present language or social communication development concerns, as reported by their parents. Parents who responded to the study advertisement were screened for eligibility with a phone call. Two children selected had been diagnosed with a language disorder and three children had parent-reported past or present concerns related to language, literacy, or communication (i.e., past selective mutism, social communication challenges, and dyslexia).

Data Collection

Data sources included an initial information-gathering phone call with the parent and a background history/demographics questionnaire. Clinical assessment tools were used to measure tic disorder severity, psychosocial functioning, core language skills, high-level language skills, narrative language skills, and social communication skills. Qualitative data included a 7-day communication log-book and a semi-structured parent interview. Children met with the examiner for two hours with one to two breaks as needed in order to complete language testing. Parents completed questionnaires during and after the assessment and diary entries were completed after the assessment.

Tools

History/ demographics Questionnaire. The following history/demographic information was collected: test scores of past language and cognitive testing, goals of past language intervention, who diagnosed the child with TS, age at TS diagnosis, current age, sex and gender, co-occurring conditions, languages spoken in the home, languages spoken at school, and parent education.

The Global Tic Rating Scale (GTRS). The GTRS (Gadow & Paolicelli, 1986) measures the severity of the child's tic disorder using four questions about how noticeable the tics are to others, how embarrassing they are for the child, how much they affect school and home functioning, and to what degree they cause social rejection (0-1 = low; 2 = medium; 3 = high). This measure was selected for its brevity and ability to group children into three groups based on severity.

The Strengths and Difficulties Questionnaire (SDQ). The SDQ (Goodman et al., 1997) is a measure of overall psychosocial functioning in children. It provides a total score for psychosocial difficulties using 25 questions about emotional symptoms, conduct problems, hyperactivity/ inattention, and peer relationship problems. Total scores are divided into four ranked categories (0-13 = 'close to average;' 14-16 = 'slightly raised;' 17-19 = 'high;' 20-40 = 'very high'), where higher scores correspond to higher levels of difficulty. The SDQ has validity and reliability evidence (Goodman et al., 1997; 2000; 2004)

The CELF-5 Core Language Subtests. The CELF-5 (Wiig et al., 2013) core language subtests create a Core Language Score that reflects general language skills. The subtests test the child's ability to select words that are semantically related, formulate sentences about pictures, repeat sentences word-for-word, and understand paragraphs. To derive a core language score,

children must complete three subtests (specific subtests vary based on their age). Percentiles and scaled/index scores are derived from age-based norms. The Core Language Score has validity and reliability evidence and it provides excellent sensitivity and specificity for differentiating between children with language disorders and typical language development (Wiig et al., 2013).

Test of Narrative Language, Second Edition (TNL-2). The TNL-2 (Gillam & Pearson, 2017) assesses story comprehension and story production. The Narrative Comprehension portion tests the child's ability to answer comprehension questions about three stories and the Narrative Production portion tests the child's ability to retell a story, generate a story about a series of pictures, and generate a story from a single picture scene. Percentiles and scaled/index scores are derived from age-based norms. The Narrative Language Ability Index has validity and reliability evidence and it provides excellent sensitivity and specificity for differentiating between children with developmental language disorder and typical language development (Gillam & Pearson, 2017).

Comprehensive Assessment of Spoken Language, Second Edition (CASL-2) Supralinguistic Subtests. The CASL-2 (Carrow-Woolfolk, 2017) measures high-level language skills through subtests assessing the child's ability to comprehend nonliteral language, derive meaning from context in scenarios, make inferences in scenarios, and interpret multiple meanings in phrases. To derive a supralinguistic language score, children must complete three or four subtests (specific subtests vary based on their age). Percentiles and standard/index scores are derived from age-based norms. The Supralinguistics Index has validity and reliability evidence and it provides good sensitivity and specificity for differentiating between children with language disorders and typical language development (Carrow-Woolfolk, 2017).

Children's Communication Checklist, Second Edition (CCC-2). The CCC-2 (Bishop, 2003) is a 70-question questionnaire that measures 10 different domains of a child's communication from the perspective of their parent. It includes questions about speech (A), syntax (B), semantics (C), coherence (D), initiation (E), scripted language (F), context (G), nonverbal communication (H), social relations (I), and interests (J). Percentiles and scaled/composite scores are derived from age-based norms. The CCC-2 combines scales A-H to derive a General Communication Composite score; the social interaction difference index takes the difference between scales A, B, C, and D and scales E, H, I, and J; and the Social-Pragmatic Communication composite score combines initiation, context, and nonverbal communication domains (E, G and H). The General Communication Composite has validity and reliability evidence. Sensitivity ranges from low to high and specificity has been demonstrated to be adequate for differentiating between children with language disorders and typical language development (Bishop, 2003).

7-Day Parent-Created Child Communication Log Book. Parents were asked to complete a 7-day log book to provide examples of their child's communication in day-to-day situations (Appendix D). They were asked 1) to provide an example of instances when the child communicated their message successfully; 2) to provide an example of when the child had difficulties communicating; 3) to make note of what else was going on in each situation; and 4) to note the reactions or responses of others who were present. The purpose of the log book was to collect recent communication examples in day-to-day communication. Log-book examples were considered when preparing for the parent interview.

Qualitative Semi-Structured Parent Interview. Parent interviews were conducted to collect additional information about the day-to-day communication of children. Parents met the

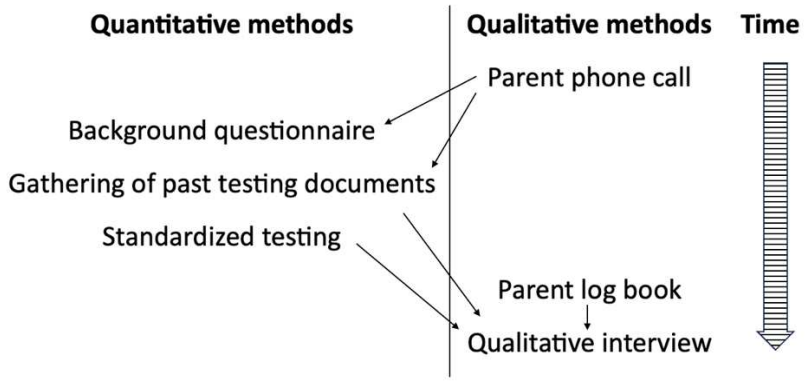
lead author in a secure video conference meeting for a 60-minute interview about their child's communication in different micro-system contexts (school, home, and peers) and to discuss barriers and facilitators to communication. Interviews were audio-recorded using a Maranz digital recorder and transcribed verbatim.

Combining information from different data sources began in the data collection phase, focusing on how information that had already been collected could inform further data collection. As cases developed, additional information was sought to create a full and consistent picture of each case. Past language and cognitive testing results were requested from all families. The general sequence of data collection and details of which information sources were used to inform others is outlined in Figure 10.

The lead author calculated scores for standardized tests and reviewed the parent diary before the parent interview. This information was used to create a tailored interview guide for each child that was based on the general protocol produced during study planning. Strengths and challenges described in the parent diary and derived from scores in testing were brought up so that the parent could describe the child's behaviour at home, expand on what happened in specific scenarios from the diary, and describe the strategies being used. The interviewer made an effort to remain open to parent perspectives that either supported or diverged from test score interpretations (consistent with the tenets of crystallization rather than triangulation; Richardson, 2000). When inconsistencies or gaps in information became evident during data analysis, parents were contacted to clarify or correct information. Data were collected between February and June 2023.

Figure 10

Mixed Methods Case Study Information Sources



Note: Arrows indicate integration between data collection processes.

Data Analysis

Data analysis was carried out by the lead author, who is a family member of a person with C-TS and a speech-language pathologist (SLP) who has provided assessment and intervention services to children with C-TS in a healthcare setting for six years. These personal and professional factors shaped the analysis process, informing and enriching the inquiry at all stages, but also requiring careful reflection on how personal and professional background/experiences influenced decisions and interpretations. The lead author made an active effort to push the boundaries of the discipline of speech-language pathology by resisting and reframing deficit-focused interpretations. Explaining how a child does better in flexible environments rather than calling the child 'rigid' or stating that a child benefits from concrete information to facilitate learning rather than calling the child 'literal' are examples of strength-based language. Additionally, children are characterized as thinking differently rather than as being deficient. We were also open to broadening the concept of communication beyond its traditional boundaries. For example, we recognized that children must be in an emotionally

regulated state to be able to communicate effectively and, therefore, we consider strategies being used to maintain emotional regulation as potential important facilitators to communication.

A four-step data analysis process outlined by Houghton et al. (2015) was used. Houghton's process is based on the work of Morse (1994), Miles & Huberman (1994) and Braun & Clark (2021; 2022). Both quantitative and qualitative information were considered at each stage. The first stage, "comprehending," was completed by transcribing qualitative interviews, engaging in data familiarization by listening to interviews and reading transcripts, memoing about initial thoughts on each case, and completing initial coding of qualitative data using Microsoft Word. The second stage, "synthesizing," involved merging information, comparing data from different sources, clarifying inconsistencies, and continually noting emerging perceptions through reflexive memos. During the synthesis stage, codes were refined and an initial summary of each case was drafted, integrating all information (background information, language testing information and qualitative data). The third stage, "theorizing," involved creating a visual map of qualitative codes for each case, noting patterns across cases (cross-case analysis), and creating a draft list of cross-case propositions from decontextualized codes. During the fourth stage, "recontextualizing," draft propositions were brought close to the data again to refine and formalize them, propositions were considered against each synthesized case, and specific examples and participant quotes were brought in to contextualize each proposition. At this stage, two to three sub-themes were developed under each proposition to further organize ideas.

Qualitative rigor was considered throughout the study using a list of strategies developed by Morse et al. (2002), including investigator responsiveness, methodological coherence, appropriate sampling, and an active analytic stance. Some examples of responsiveness included

adjusting the data collection plan and documenting decisions in an audit trail, using reflexive memos to encourage interpretations that went beyond initial impressions of the data, and carrying the bioecological model through each stage of the project while also leaving space for bottom-up developments. Methodological coherence was maintained through carefully selected data collection methods. To ensure appropriate sampling, parent-child dyads were chosen purposefully to provide a variety of examples of children with C-TS. An active analytical stance was maintained through all of the strategies mentioned thus far and by integrating theory into the research process.

Results

Case Profiles: Background information and intervention history

Tables 10 and 11 provide a summary of demographic information and testing scores from previous cognitive testing for each case. Tables 12-14 outline the results of language and communication testing measures completed for the project. All children have been given a pseudonym and some details that could identify them have been changed. A brief introduction to each case is presented below, providing background and intervention history. Day-to-day examples of how language strengths and challenges show up in different contexts and the strategies/supports being used by each family are discussed under the 'bioecology of day-to-day communication.'

Case 1: "Kent"

Kent is an eleven-year-old boy in a family of four children, all with various developmental challenges. He spoke his first words after two years of age and began combining words into very simple sentences around three years of age. Kent's mother had early concerns about Kent's language development but was reassured by the doctor that boys and first-born

children sometimes speak late. She was primarily focused on behaviour concerns with Kent when he was a toddler and a preschooler and was very busy with two other children who were subsequently born in quick succession. Kent was first referred for language assessment at the age of five just before entering school. The consulting SLP concluded that "language skills are well within normal limits;" however, no formal language testing was documented at that time. That year, the family had been accessing psychiatry for help with Kent's behaviour. Upon entering school, Kent's psychiatrist recommended that he receive extra support in his first year of school through a program that offered specialist consultant services, including speech-language pathology. Kent's mother stated, "he had a letter from the psychiatrist stating that he required funding, and should be eligible… and the school said that they chose not to access it." The following year Kent did receive funding for extra support, however, challenges specific to language still had not been formally identified. Kent was identified with a language disorder at the age of 7 years, in his third year of school, when he attended a mental health intervention program where a comprehensive assessment across various areas of development was completed. Kent was diagnosed with a "moderate expressive language delay" and "moderate receptive language delay." Kent received the support of an SLP through his school that year and the following year. Kent's mother had been working with the school each year to develop a plan that would meet Kent's other needs as well. Unfortunately, Kent's mother's experience of engaging with the school to develop and maintain accommodations for children with TS needs and other physical and learning needs was not positive. She described it as "a constant battle" and stated that, "every three months would be another fight for the same thing." At the time of data collection, Kent's language testing scores indicated continued challenges with language

development that appear consistent across the areas tested: core language, narrative language, supralinguistics, and social-pragmatics (Tables 12-14).

During the COVID-19 pandemic, the family was forced to learn at home for an extended and Kent's mother discovered the of removing the traditional school setting from Kent's day-to-day life and learning environment. She implemented accommodations at home that the school had not done successfully. For example, she would allow Kent to start his day off with active outdoor play, like bike riding, and he would complete focused school work later in the day. At the time of the parent interview, Kent was in his second year of home-schooling and was receiving in-home services from an SLP, an occupational therapist, and a behaviour consultant through a combination of disability funding and home-school funding. He was receiving medical support through psychiatry.

Case 2: " Cam"

Cam is an eight-year-old boy in a family of four children, all with various developmental challenges. There were no concerns about Cam's language and motor developmental milestones; he spoke his first word at approximately one year of age and combined words at approximately two years of age. Cam's mother noted some concerns with behaviour in the toddler and preschool years and chose to access psychiatry support before Cam entered school. In his first year of school, Cam's mother believed that "he's struggling more than he should be." She was concerned about his peer interactions and comprehension in the classroom. The COVID-19 pandemic interrupted Cam's first year of schooling and ultimately led to a decision to shift to home-schooling. Cam's language skills were first tested when he was 7 years of age. The assessment consisted of the CELF-5 Screening Test (Wiig et al., 2013); test scores were not reported. The report concluded that "[Cam] did not reach criterion on the screening test,

suggesting that further evaluation was recommended." Following this assessment, Cam received intervention from an SLP. At the time of data collection, Cam's language testing scores indicated continued challenges with language development with relative strength in core language (as measured on the CELF-5). Areas of challenge included narrative language, supralinguistics, and social-pragmatics. At the time of the parent interview, he was receiving support services (speech-language pathology and behaviour consultation) through a combination of disability funding and home-school funding. He was also receiving medical support through psychiatry.

Case 3: "Jasper"

Jasper is a nine-year-old boy in a family of three children, all with various developmental challenges. Jasper spoke his first words at approximately ten months of age and began combining words around 14 months. As a preschooler, Jasper experienced a speech sound delay and exhibited selective mutism. He received speech-language pathology service during the year prior to entering school and during his first year in school, and no longer exhibits either of these challenges. Jasper's language skills were evaluated at 4 years of age and were measured within normal limits on the Clinical Evaluation of Language Fundamentals, Preschool, Second Edition. A relative strength in receptive language was noted (receptive language was high average and expressive language was average). Despite overcoming selective mutism, Jasper's mother said that he continued to sometimes have trouble his ideas. At the time of data collection, Jasper's test scores were consistent with age-expected language development in the areas of core language, narrative language, and supralinguistics. His only area of apparent difficulty was Recalling Sentences on the CELF-5. The previous pattern noted by his SLP is evident again in Jasper's TNL-2 scores, with the receptive language portion (Narrative Comprehension; average) a relative strength compared to the expressive language portion (Narrative Production; low

average). Jasper's social-pragmatic language skills were measured as below average. Jasper attended a French immersion school program for the first four years of his schooling and moved to a program for gifted students in the current year with classroom accommodations in place. At the time of the parent interview, Jasper was receiving medical support through psychiatry and neurology.

Case 4: "Lorenzo"

Lorenzo is a ten-and-a-half-year-old boy in a family of three children, all with various developmental challenges. Lorenzo spoke his first word around 11 months and began combining words at 14 months of age. Lorenzo's parents did not have any current or past concerns about speech and language development and Lorenzo had not had any assessment or intervention with an SLP; however, Lorenzo's mother noted some challenges with social communication, such as, "knowing his audience." At the time of data collection, Lorenzo's test scores were consistent with age-expected language development in narrative language and supralinguistics. His scores on the CELF-5 indicated advanced core language skills. Lorenzo's social-pragmatic language skills were measured as below average. Lorenzo attends a school program for gifted students with classroom accommodations in place. He receives medical support through psychiatry and neurology.

Case 5: "Mora"

Mora is an 11-year-old girl and an only child. Mora spoke her first word before the age of one and began combining words before she turned two. Mora's parents did not have any current or past concerns about speech and language development and Mora has not had any assessment or intervention with an SLP. Mora had some challenges with reading and writing development in her early school years. In her fourth year of school, she participated in a psychoeducational

assessment and was identified with dyslexia. Mora received intensive intervention for literacy beginning that year through her school and her literacy skills were brought up to grade level. At the time of data collection, Mora's test scores were consistent with age-expected language development in the areas of core language, narrative language, supralinguistics, and social-pragmatics. Her only area of apparent difficulty was Recalling Sentences on the CELF-5. Mora was attending a traditional school classroom with access to accommodations for her Tourette and ADHD symptoms at the time of the study. Mora was receiving medical support from a neurologist.

Table 10

Case Demographic Information

	Sex	Age (yrs)	Age of TS diagnosis	Tic disorder severity	Total psychosocial difficulties	Co-occurring conditions	Languages in the home	School language exposure	Maternal education
Case 1 "Kent"	M	11	6	High	Very high	ADHD; Learning disability; Depression; Language disorder; periodic limb movement disorder; suspected Anxiety disorder; delayed early motor milestones	English	English	College degree
Case 2 "Cam"	M	8	6	High	Very high	ADHD; Learning disability; Depression; Language disorder	English	English	College degree
Case 3 "Jasper"	M	9	6	Medium	Slightly raised	ADHD; Anxiety disorder; Past selective mutism; Past speech sound delay	English	English/ French	Graduate degree
Case 4 "Lorenzo"	M	10.5	7	Medium	Very high	ADHD; OCD; Anxiety disorder	English	English	Graduate degree
Case 5 "Mora"	F	11	8	Medium	Average (no difficulties)	ADHD; learning disability (dyslexia)	English	English/ French	Bachelor's degree

Table 11

Past Intellectual Testing Scores

	Intellectual Functioning on the Wechsler Intelligence Scale for Children® Fifth Edition (WISC-V) Description (Composite Score)						
	Age at assessment (yrs)	Verbal comprehension	Visual spatial	Fluid reasoning	Working Memory	Processing speed	Full Scale IQ
Case 1 "Kent"	9.5	Extremely low (64)	Average (98)	Low average (88)	Very low (72)	High average (112)	Not reported[a]
Case 2 "Cam"	7	Very low (75)	Average (89)	Average (88)	Low average (80)	Average (89)	Low average[b] (81)
Case 3 "Jasper"	7.5	Very high (121)	Average (109)	Very high (127)	Average (116)	Very high (121)	Very high (123)
Case 4 "Lorenzo"	7	Extremely high (130)	High average (112)	Very high (121)	High average (119)	Very high (121)	Very high (129)
Case 5 "Mora"	7.5	Very high (121)	Average (104)	Average (94)	Low average (86)	Average (109)	Average[b] (104)

[a]Full scale score not reported due to discrepancies between composite scores.

[b]Consider with caution due to discrepancies between composite scores.

Note: < 69 – Extremely low; 70-79 – Very low; 80-89 – Low average; 90-109 – Average; 110-119 – High average; 120-129 – Very high; > 130 – Extremely high.

Table 12

CELF-5 Scaled and Standard Scores

	Clinical Evaluation of Language Fundamentals, 5th Edition (CELF-5) Scaled/Standard Score (Percentile) [95% Confidence Interval]						
	Word classes *M* = 7-13 *SD* = 3	Sentence Comprehension *M* = 7-13 *SD* = 3	Word Structure *M* = 7-13 *SD* = 3	Formulated Sentences *M* = 7-13 *SD* = 3	Recalling Sentences *M* = 7-13 *SD* = 3	Semantic Relationships *M* = 7-13 *SD* = 3	Core Language Score *M* = 85-115 *SD* = 15
Case 1 "Kent"	8 (25) [6-10]	-	-	5 (5) [3-7]	4 (2) [2-6]	5 (5) [3-7]	75 (5) [68-82]
Case 2 "Cam"	-	12 (64) [10-14]	6 (9) [4-8]	9 (37) [7-11]	9 (37) [8-10]	-	93 (32) [86-100]
Case 3 "Jasper"	-	11 (63) [9-13]	8 (25) [6-10]	16 (98) [14-18]	6 (9) [4-8]	-	101 (53)[a] [94-108]
Case 4 "Lorenzo"	18 (99.9) [16-20]	-	-	10 (50) [8-12]	14 (90) [13-15]	10 (50) [8-12]	117 (87) [111-123]
Case 5 "Mora"	9 (37) [7-11]	-	-	10 (50) [8-12]	5 (5) [3-7]	10 (50) [8-12]	90 (25)[a] [83-97]

[a]Interpret Core Language Score with caution due to variability across subtests.

Note: Recommended Core Language Score cut-off for language disorder = 80

Table 13

TNL-2 and CASL-2 Scaled, Standard, and Index Scores

	Test of Narrative Language, 2nd Edition (TNL-2) Scaled/ Index Score (Percentile) [Standard Error of Measurement]			Comprehensive Assessment of Spoken Language, 2nd Edition (CASL-2) Standard/ Index Score (Percentile) [95% Confidence Interval]				
	Narrative Comprehension *M* = 7-13 *SD* = 3	Narrative Production *M* = 7-13 *SD* = 3	Narrative Language Ability Index *M* = 85-115 *SD* = 15	Nonliteral Language *M* = 85-115 *SD* = 15	Meaning from Context *M* = 85-115 *SD* = 15	Inference *M* = 85-115 *SD* = 15	Double Meaning *M* = 85-115 *SD* = 15	Supralinguistic Index *M* = 85-115 *SD* = 15
Case 1 "Kent"	6 (9) [4-8]	7 (16) [6-8]	80 (9) [74-86]	82 (12)	87 (19)	79 (8)	81 (10)	80 (9)
Case 2 "Cam"	6 (9) [5-7]	4 (2) [3-5]	72 (3) [68-76]	70 (12)	75 (5)	76 (5)	-	72 (3)
Case 3 "Jasper"	11 (63) [10-12]	8 (25) [7-9]	94 (35) [90-98]	114 (82)	119 (90)	111 (77)	-	114 (82)
Case 4 "Lorenzo"	12 (75) [10-14]	11 (63) [10-12]	108 (70) [103-113]	109 (73) [106-112]	125 (95) [121-129]	117 (87) [114-120]	107 (68) [103-111]	114 (82) [112-116]
Case 5 "Mora"	15 (95) [13-17]	10 (50) [9-11]	113 (81) [107-119]	107 (68) [64-72]	110 (75) [71-79]	110 (75) [72-78]	103 (58) [53-63]	107 (68) [105-109]

Note: Recommended Narrative Language Ability Index cut-off for language disorder = 92; Recommended Supralinguistics Index cut-off score for language disorder = 85.

Table 14

CCC-2 Scaled and Composite Scores

	Children's Communication Checklist, Second Edition (CCC-2) Scaled/Composite Score (Percentile) [95% Confidence Interval]									
	Speech *M* = 7-13 *SD* = 3	Syntax *M* = 7-13 *SD* = 3	Semantics *M* = 7-13 *SD* = 3	Coherence *M* = 7-13 *SD* = 3	Initiation *M* = 7-13 *SD* = 3	Scripted language *M* = 7-13 *SD* = 3	Context *M* = 7-13 *SD* = 3	Nonverbal Communi-cation *M* = 7-13 *SD* = 3	Social relations *M* = 7-13 *SD* = 3	Interests *M* = 7-13 *SD* = 3
Case 1 "Kent"	6 (9)	2 (0.4)	4 (2)	3 (1)	7 (16)	6 (9)	2 (0.4)	9 (37)	4 (2)	2 (2)
Case 2 "Cam"	3 (1)	1 (0.1)	5 (5)	4 (2)	7 (16)	5 (5)	3 (1)	8 (25)	4 (2)	7 (16)
Case 3 "Jasper"	9 (37)	12 (75)	11 (63)	8 (25)	16 (98)	3 (1)	9 (37)	5 (5)	5 (5)	7 (16)
Case 4 "Lorenzo"	9 (37)	12 (75)	11 (63)	8 (25)	3 (1)	4 (2)	4 (2)	8 (25)	8 (25)	2 (0.4)
Case 5 "Mora"	12 (75)	12 (75)	10 (50)	9 (37)	10 (50)	10 (50)	10 (50)	10 (50)	10 (50)	16 (98)

	General Communication Composite *M* = 85-115 *SD* = 15	Social Interaction Difference Index	Social-Pragmatic Communication Composite
Case 1 "Kent"	70 (2)	+9	85 (16)
Case 2 "Cam"	67 (1)	+13	75 (5)
Case 3 "Jasper"	86 (18)	-17	75 (5)
Case 4 "Lorenzo"	79 (8)	-19	75 (5)
Case 5 "Mora"	103 (58)	+3	100 (50)

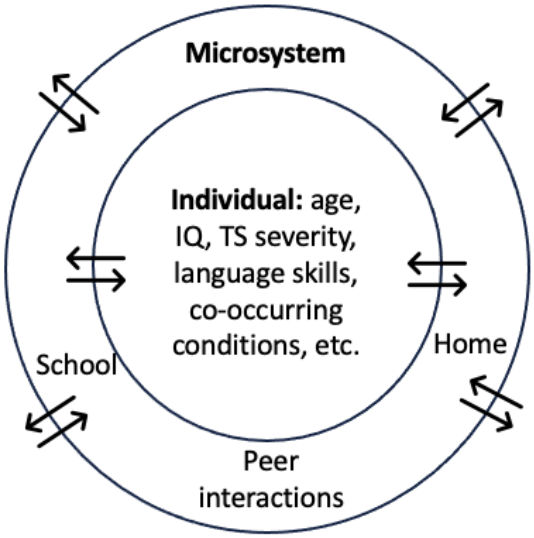
Note: Recommended General Communication Composite cut-off for language disorder = 85; A Social Interaction Difference Index of < -11 indicates social interaction challenges.

The Bioecology of Day-to-Day Communication

The bioecological model (Figure 11) characterizes the complex interaction between child characteristics and the various settings in their micro-system. It is a visual representation of how an individual's characteristics (e.g., diagnosis, language ability, and cognitive profile) interact with factors in various day-to-day contexts to explain how development occurs. The parents of the five children in this study were all implementing several flexible strategies across their education, home, and social settings to meet various needs related broadly to their child's day-to-day communication (including language learning, learning more generally, emotional regulation, family interaction, and social success).

Figure 11

Bioecology of Day-to-Day Communication in Children with C-TS



Proposition 1: The Learning Setting Requires Careful Consideration

Unique learning needs sometimes interacted in complex ways with TS symptoms, language and communication, and symptoms of other co-occurring conditions. We discuss this under two sub-themes: modified school programming and success with accommodations.

Modified School Programming

Kent and Cam were both experiencing significant problems in the traditional in-school learning setting. Kent's parents had tried to work with his school over multiple school years in order to set things up so he could be successful. These families decided to shift to homeschooling for their children and succeeded with this option. Prior to this change, Kent's mother described observing self-harm behaviours due to his inability to cope with the in-school setting:

> He would get off the bus, have a rage attack, and be in bed by 4:30. He'd miss dinner, he'd miss everything… It was not doing him any favours, considering we're already making all these accommodations, continually, for his educational needs, and really he's not gaining in any aspect, and he's got no life outside of school because he can't handle it. He's suppressed all day long [at school], that now he's, really, I mean, we're about at a padded bedroom and an iPad, that's where we're heading. He's got no management of anything.

For Cam, there was a lot of irritability occurring due to sensory sensitivities in school combined with not understanding what was going on in the classroom, especially when it came to communicating with peers. Cam's mother describes, "he's sitting there and smiling and complying. He's not really getting anything; he's just suppressing himself to blend in and, 'please, dear God, don't ask me questions.'" "[There was] no benefit for keeping him in if he's not understanding. You're frustrating him and frustrating the peers." For these families, the success of homeschooling has come from the flexibility it provides, the ability to integrate self-care and adaptive skills into their daily goals, and the ability to target learning areas that need additional focus, particularly language skills. For Kent, the flexibility of homeschooling allowed him to meet his high need for physical movement for self-regulation. His mother focuses largely

on language and communication-related skills as she sees their practical importance for future success. She describes her approach:

> Just making that accommodation, and understanding that…these are things you should know, and just really honing in on the things that are important. So photosynthesis, really, in the realm of what you're going to be doing is not important. If you can't understand, just by my tone, whether I'm happy or mad, that's going to affect you… [Kent's] big thing is we work on comprehension. He hates it. It's ongoing. Lots of reading, trying to decide what's going to happen next. Sequencing is a big, big issue.

Kent's mother likened the discovery of at-home learning to an "enlightenment." She also discussed the benefits of in-home support for language and other areas of development that she was not able to access regularly when Kent was learning in the classroom.

> We're a slow and steady family. There is no rush to the finish line, we're in no race. It's really just keeping them engaged and keeping the supports there, keeping [Kent] conscious and aware of all those things. If you're always engaged in speech and language development, it's just something that you are expected to do."

Kent's mother applauded the in-home support team that she works with and described the flexible approach that she and the consultants take with Kent.

> You can't have expectations on the language. When you're communicating, you need to know when to pry for more information, and realize that it's just not their day today. They're having a hard enough time

> controlling their tics to even talk to you without freaking out. So, just understanding not every day is the same for these Tourette kids. So, today I perform well, tomorrow I can't perform at all, the next day I'm amazing, and the next day I'm mediocre. Every day is different, and you have to just roll with that instead of, well, if every day is mediocre it'd be okay. Every day is not mediocre. One day [Kent's] ticking so bad that we are not learning any new concepts. We are just trying to stay on task for a half an hour, and then other days he's not ticking, and 'alright, I'm going to just get her done.' 'Okay, let's get her done', and so I'll push him and add more work on those days. But it's about flexibility, and understanding they're not doing it to themselves; they're not doing it to you. They're as annoyed as you are.

Jasper and Lorenzo's learning needs differ from those of Kent and Cam. They both attend a special program designed for children who have above-average intellectual functioning. Both boys benefited from accommodations and special programming to enhance learning and school functioning. Both boys have more flexibility with their school attendance than is typical. For Lorenzo, his mother stated, "there's been days where he's like, 'Mom, I can't go to school today. Like, I don't have the capacity to go,' and we'll typically respect that, because otherwise I just get a call to pick him up anyways." This flexibility can mean staying home, but also sometimes involves being picked up early. Jasper's mother shared an example of being called by the school to pick him up: "I got a call from the principal asking to pick him up because, so, she didn't know what precipitated it, but he just was not listening and was not going to stay in the

classroom, and he kept just trying to leave." Jasper's mother believes that he was struggling on that particular day with an unexpected transition to a new teacher, but that he was not able to express what was upsetting him.

Both of the boys are often given a separate quiet area in the school to do their work, away from the other students. Jasper's rambunctious and constant vocalizing can sometimes disrupt classroom learning in school (e.g., standing on a chair singing during class time) and his mother stated "you could see reflected in his work what kind of day he'd had. So, you know, he would have a math test and he would miss half the answers, and he's good at math." For Lorenzo, his mother described that his tendency to hold the adults at the school to a very high moral standard, question their decision-making, and be very frank about his views was not always received well.

Success with Accommodations

Mora was finding success with some specific supports and accommodations in a regular school program. Her mother reported that they have had "phenomenal" support from their school regarding Mora's needs. Mora's challenges with literacy learning were quickly identified, she was provided with testing to receive a dyslexia diagnosis, and intervention followed. She is now doing well with literacy in school. Her mother shared an example of the support that Mora has gotten for her tics in the classroom:

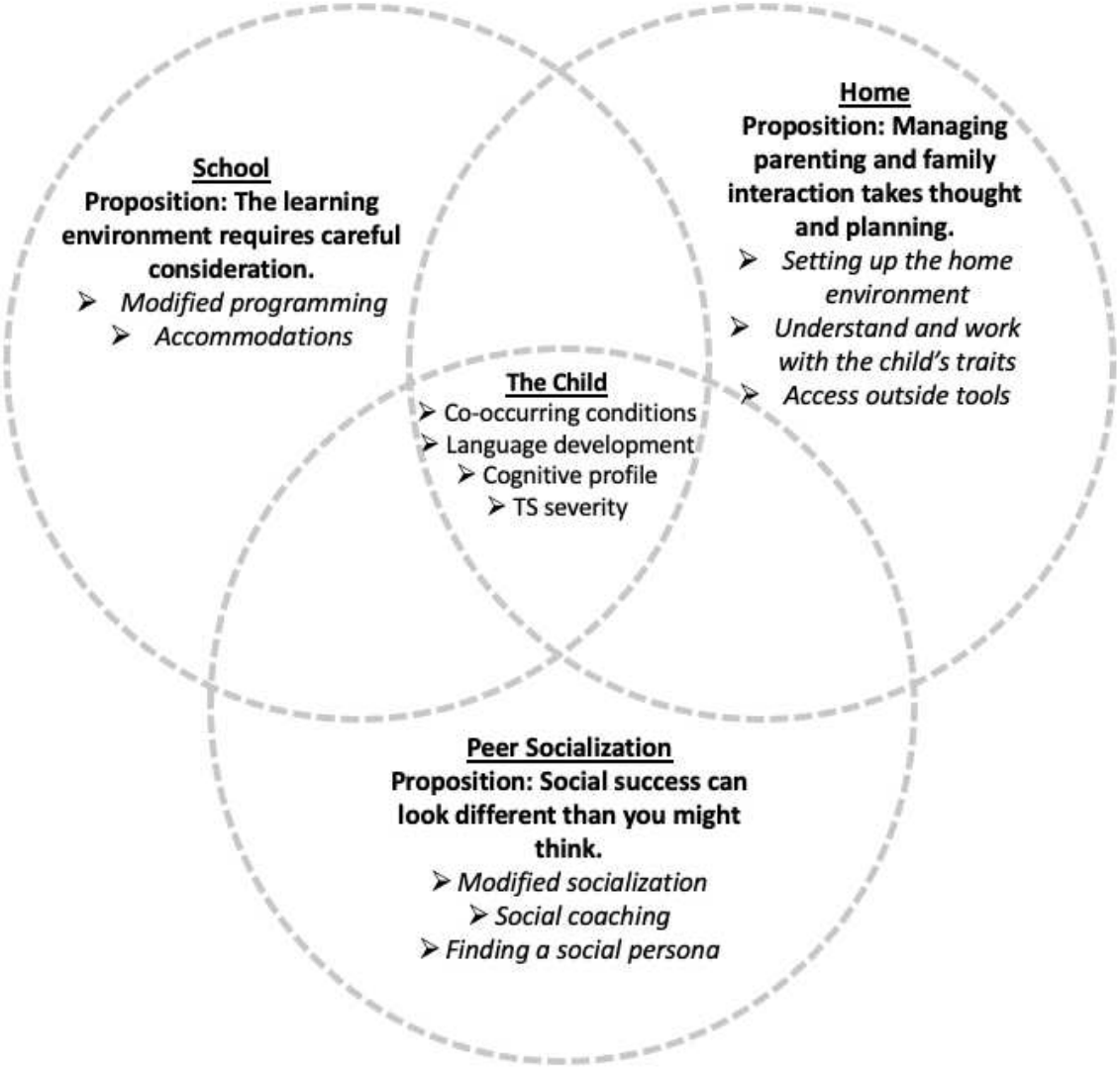
> The teacher pulled her aside and said, '[Mora], I noticed that you were ticking a bit. Is it something that I'm doing or anything that I can help you with?' [Mora's] like, 'no, it's okay, it just happens.' So, yeah, she has a lot of support."

Mora also has other accommodations available to her, such as using a quiet space

for exams, speech-to-text, and text-to-speech; however, her mother reported that she does not always choose to use these tools. "I'd asked her last year, 'Why don't you use it?' and she said, 'Well, I want to challenge myself so that I know how to write, and be able to do it on my own without a computer helping me.'" Mora's mother also discussed the positive effect of having a TS expert visit the classroom to educate the students and teachers about TS and what it's like to live with tics.

Figure 12

Bioecology of Day-to-Day Communication: Propositions and Subthemes



Proposition 2: Managing Parenting and Family Interaction Takes Thought and Planning

All parents put a lot of thought into how they manage family interactions, shape the home environment, strive to understand their child, and access the support of outside tools. We discuss this under three sub-themes: setting up the home environment, striving to work with and understand the child's traits, and the use of outside tools.

Setting up the Home Environment

Parents who have more than one child with neurodevelopmental conditions or learning challenges in their households discussed how they manage family dynamics between siblings, especially where Tourette/ADHD behaviour, sensory differences, and other personality traits intersect. In Cam's household, where there are four children with learning needs, the household dynamic was described as "chaotic" and his mother described how she creates structure by giving each child specific responsibilities and rewarding them when they have completed these tasks. The children are highly motivated by screen time and this is a primary reward for them. Cam's mother discussed her natural tendency to be a more 'laid back' parent, but how the nature of the household requires her to be stricter. "In another life, I was a great mom and super lax and super easygoing, but that's not the children I have so that's not the mom I can be."

For both Kent and Cam, socially inappropriate tics come out most at home and can be disruptive to other family members (e.g., crude gestures, spitting, swearing). Kent can become aggressive when triggered and his family had implemented a "no play fighting" rule because things can get too rough very quickly. For Cam, he could easily enter an emotionally dysregulated state, "just crying, whining, hyper, just not quite regulated," and this disrupted other family members. Cam's mother described how engaging with him physically in a playful way can support him when he's getting overstimulated:

> When he's dysregulated, he will tic a lot more, he will bug and pester, and he will keep amping up, amping up, amping up until he either has a huge meltdown and ends up in restraint or until he's able to engage in something else to release that energy. So, he doesn't like being tickled, and yet he likes being tickled, so sometimes it would be like, 'Do you want a tickle, buddy? That's it, I'm going to tickle you,' and so I'll lay on him, and pin his arms down, and pretend to tickle him. Not a hard tickle, because that will spark him into a rage, but a tease tickle.

Another strategy that parents discussed was the importance of quiet moments and space away from siblings. For Kent, his mother spends time cuddling him and uses it as an opportunity to talk through conflicts that came up earlier, but that Kent was not able to discuss calmly at the time.

Jasper brings a lot of energy to their home and his mother described what they do to manage the busyness and overstimulation that can come from having three children in the house:

> Sometimes it's hard because it's hard to focus him back. So, you're like, 'Okay, [Jasper], it's time for supper' or whatever, and he's still, like, ['specific repetitive singing tic'], and you're like, 'okay, I know you like that song, but we need to focus on what we're doing.' If he's doing that over top of another sibling who's trying to tell us something, so, if we're sitting at the table, and we're like, 'How was school today?' we're talking to [sibling], and then he'll interject over top and then it annoys his siblings, then they're shouting at him to stop, which doesn't make him stop, and then he usually just gets louder.

To give everyone a break, the family creates "zones" in the house where each child can read or do another quiet activity. Jasper needs a high level of stimulation when he is on his own,

so he spends his quiet time listening to an audiobook while he does a craft or something else creative. His mother described him as a "sensory seeker," he even prefers to sleep with the lights on and music playing.

Lorenzo is quite sensitive to noise and even though he is the oldest child, he gets picked on because his siblings know what to say to trigger him. Noise and teasing can cause him to become overstimulated and/or overwhelmed and he may digress into "baby talk," where, "he just kind of shuts down in terms of talking, one word sentences, or he's very whiny, baby sounding." His mother provided an example:

> Just two days ago the kids were at the park. [Lorenzo] fell at the park, I guess, and the kids kind of laughed because he fell, but it wasn't a big fall, but something about that event was really upsetting. He came home, he laid on the couch, he put a blanket over his head, and he was just like, 'No, not talk, not talk,' and it's such a departure from his typical behaviour and language that it's really weird to see when he's like that.

In these examples, Lorenzo sometimes needs time before he's able to accept any help, but sending the family's dog to his room can be helpful, "just petting the dog tends to really help." If he's open to it, his mother will lay with him and rub his back in a quiet low-lit space.

Like other families, Mora's mother shared the importance of quiet moments. She finds that Mora is on the quiet side personality-wise, but her communication skills are excellent in most settings. When she is feeling highly anxious, she will not talk and she has experienced some periods of mutism at school associated with the stress of the early COVID-19 pandemic. However, in the absence of major stressors, she can engage with others easily and express herself in a variety of environments. Mora will share very little with her mother, however, when asked

what she's been up to or about her day. "She hates me asking questions, like *hates* it." Mora's mother has found that asking about her day right when she is picked up doesn't go well, but when they are more relaxed, Mora tends to share openly.

> I'll be putting her to bed or whatever, and we'll just be laying together, and she'll just be like, blah, and tell me all the things, and it just warms my heart so much, and I love it when she does that, so I try and make those situations as much as possible.

Mora's mother finds that connecting at night time when things are more relaxed tends to help Mora share about her life.

Kent's mother discussed modifying the environment to support communication as well. She sees that Kent has limitations in terms of how much communication burden he can handle in a day. She sets things up to give him breaks when she knows that there will be a lot of talking and listening expected in a day, "I think that he's very dependent on me in his communication to set him up with minimal words because I know we're going somewhere where he's going to be talked to and it's going to be more draining for him, so less engagement."

The mothers of the four boys discussed many behaviour strategies used at home to set their children up for success. All households use a high level of structure and routine, but with built-in flexibility. Parents also mentioned the use of consequences when appropriate. Jasper and Lorenzo's parents mentioned the importance of ignoring socially inappropriate tics, to not reinforce them.

Striving to Understand and Work with the Child's Traits

A few parents discussed how they tend to accept and work with their children's personality characteristics rather than fighting them or trying to change them. A longstanding

characteristic for Kent is that he has trouble contending with ambiguity; he does best when information is concrete, “there’s black and there’s white, no in between.” Kent’s mother accepts this characteristic. “He is who he is. There’s nothing I’m going to be able to do to beat it out of him. That’s him, and my job is to try to work with that personality.” Similarly, Lorenzo has some unique personality traits; he is highly committed to justice and fairness, is vocal in sharing his views and knowledge, and will disagree with and challenge others directly, regardless of who they are. His mother expressed that these qualities and behaviours are understandable, as she also had these qualities as a young person. She tends to coach him gently, rather than reprimanding him, “I think I have more tolerance for it because I get it (laughs) because I was sort of like that, so I just will be like, ‘just try not to interrupt her today,’ but I’m not like, ‘you can’t talk to your teacher like that.’ Lorenzo’s mother also expressed that she did not know social communication was an area of communication development for which SLPs could provide support.

Mora’s mother mentioned a desire to improve her understanding of her daughter so that she can be the best parent possible for her:

> I would like to be educated enough that I can understand where she’s coming from too. I always just make sure that we communicate as best we can because we have such a short time together, so, you know, making this the best it can be while we’ve got it… for her Tourette’s we have had supports and resources, but ADHD, not really. So, I’m still learning as well. So, I just try and go with what she says, you know, what works for her, what she asks for, and then I try and be accommodating.

Outside Tools

Parents were taking an active role in choosing and using outside tools and services, including medications, consultants (SLP, OT and behaviour consultants), doctors (psychiatry and neurology), and technology. Kent and Cam's parents had advocated hard for the help of consultants at various points, sometimes paying for assessments out of pocket to demonstrate the boys' needs and also seeking intervention from private practitioners as required. A few parents mentioned medication as a tool for managing symptoms of TS, ADHD, and mood problems.

Parents were taking an active role by documenting side effects of medications and working with their doctors to ensure that medications were helping. For Cam, a medication that was tried recently for managing his mood needed to be discontinued because it caused insomnia, which limited his ability to function during the day. For Jasper, ADHD medication supports him with listening and getting tasks done throughout the middle of the day, when it is most active in his system, but, "mornings are tough for him." Mora's mother reported that they had decided to change providers when the doctor would not listen to Mora's preferences around her medication. Her mother reported that "[the medication] just made her feel really crummy." She explained, "So basically, he denied her choice of her own body and that freaked her out, so we found another doctor because he wouldn't lower our meds for us."

In the homes of Kent, Cam, Jasper, and Lorenzo, screen time is often used as a tool to provide rewards for accomplishing tasks and as downtime. Kent's mother has begun to let him use the verbal 'chat' function on his Fortnight video game, despite her concerns about the risks associated with letting him chat with strangers. She has found that Kent, who is generally not very motivated to verbalize his thoughts, puts in extra effort to communicate clearly with his teammates and to collaborate on a game strategy. His mother has been impressed by how much

language expression practice Kent gets through chatting with his teammates. She is careful to check in often to make sure that the discussion taking place during gameplay is appropriate.

Both Kent and Lorenzo's mothers mentioned the importance of recognizing needs and seeking support for 'communication' development, rather than focusing solely on articulation or "[being] stuck in the speech, 'do you have a lisp? Can you pronounce your letters?'" (Kent's mother).

Proposition 3: Social Success can Look Different than You might Think

Several specific strategies and supports were being used by the parents regarding socialization. Parents of all five children expressed that their child had found success socially; however, Kent, Cam and Lorenzo's mothers described specific hurdles and the need for assistance in their journeys to social success. Jasper and Mora were being provided with minimal help socially. We discuss this under three subthemes: modified socialization, social coaching, and finding a social persona.

Modified Socialization

Both Kent and Cam had struggled with socialization at school when they were attending. These parents described modifying the socialization environment to allow the children to be successful. Kent's mother explained that the shift to homeschooling had had a very positive impact on his social life. His mother described:

> We have never had friends outside of our school, and even those [school] relationships were really, really strained. Just a lot of impulse control and stuff like that, that goes along with ADHD and Tourette… We weren't invited to their birthday parties; we weren't engaged with them in any way…but now he's got

friends outside school. So, when they get home, he's out running around with friends. He's got community involvement.

Now that the family is more active in the community, Kent has become friends with another boy who also has TS. They had previously gone to the same school but had not known each other. The friendship between the boys works very well because both boys have periods where they are struggling with their tics and there is an understanding in the friendship that they sometimes won't see each other for an extended period. "They hang out basically every third day for a long time, and then they won't see each other for, like, three months." They always come back to the friendship when they can.

Cam's family participates in community activities through the homeschool program, like skiing, where Cam often has social opportunities through meeting new children who are also participating that day. Cam benefits from the opportunity to make new friends who he can have a positive interaction with for the duration of the activity.

> That means they have one day where they have a friend, and then we don't have to see that person again. We don't have to account for the fact that the N word is a new tic. We don't have to revisit that and go through the remorse and the 'I'm sorry' and 'Hey, something's wrong with me.'

Cam's mother explained that he can use suppression of his tics to mask during interactions outside of the family. She discussed the positive benefits of this strategy for socialization, "he gets it out within the context of his siblings, in a safe environment, and then is able to control it and go out and make good relationships." Kent also has tended to rely on masking and suppression, "he has mastered the skill of being able to tic really quite a bit and then cut it right off, down to nothing." The negative aspects of masking Tourette symptoms

through suppression were touched on as well, for instance, attempting to suppress all day long while learning in a classroom led to exhaustion and an inability to regulate emotions in the evenings.

Social Coaching

A few parents described coaching their children in social communication skills. Mora's mother attributes Mora's positive social communication skills in part to the coaching that she has provided. "I always just try and teach her that the way you speak to somebody, and the tone that you use, and being honest, is always the best way to deal with anything, whether it's uncomfortable or not." Mora's mother also discussed the importance of letting one's needs be known but in a respectful way. "You need to tell people too. They can't read your mind. If things aren't going your way or if you're upset, you have to tell them, but always try and tell them gently."

Lorenzo's mother also discussed providing social coaching. As an example, Lorenzo became very upset when a close friend was almost seriously hurt during one of his activities. Lorenzo wanted to insist that his friend give up the activity. Lorenzo's mother supportively encouraged him to not get involved by gently telling him, "I know that you're concerned about him, but it's not our place to tell somebody that they can and can't do something." When he is upset with a decision made by an adult, he benefits from an explanation of the adult's thought process and reasoning. In his class, different students tend to have different expectations and rules that they do or do not need to follow. His mother finds that discussions of 'equity' versus 'equality' can support him in accepting that different students are treated differently by teachers.

Lorenzo attends an informal social group for children where they meet online to play video games. It's been a great opportunity for Lorenzo to take on some social leadership: the

young adult who moderates the group sees Lorenzo as a competent leader of the group and will leave him to moderate if the kids want to continue playing after the group time is up. Lorenzo is often perceived by adults as competent beyond his years because of his sophisticated vocabulary and advanced knowledge in many subjects. However, Lorenzo's mother does sometimes worry that he could be left with responsibilities that go beyond his abilities, "he is like talking to a small adult sometimes, and it is hard to remember that he is not an adult."

Finding a Social Persona

Although Jasper's constant verbalization was described as a challenge in the home environment, his mother described how this silly persona seemed to make him well-known and well-liked by his peers, "kids know who he is, and I would say they look at him favourably." Regarding Jasper's impulsive behaviour at school, like a recent example of him standing on a chair singing in the classroom, his mother explained, "I think eight-year-old boys think that kind of stuff is funny, so it's not really been a social problem for him." Jasper was attending a new school and had already developed some good friends in the first part of the year. He had found a way to make himself well-known and liked while being his rambunctious self.

Discussion

This study aimed to collect information related to language and communication development in five cases of children with C-TS in order to increase our understanding of communication development in C-TS. Using the bioecological perspective, children's unique personal profiles (e.g., diagnosis, language ability, and cognitive skills) and their day-to-day communication were considered in order to identify some of the communication barriers and facilitators present in the school, home, and peer socialization contexts.

These five cases speak to the variety of profiles that can exist in the C-TS population. There was variation across cases in terms of cognitive profiles, severity of tics, core language skills, social challenges, and the overall level of support needed. The children also had variable school experiences: while Mora's difficulties had not had a great impact and she had received help at school where she needed it, Kent demonstrated significant challenges across areas of development and had had negative experiences with finding a supportive school environment. The parents in this study implemented a variety of unique strategies to scaffold their children's communication in the home context. All parents discussed elements of needing to foster a calm environment and encourage emotional regulation to facilitate communication. The needs for support with social development varied as well. While some children appeared to be receiving minimal assistance, others received help from parents and through accessing services outside the home. This variability is not surprising, given that past research indicates two-thirds of children C-TS experience problems relating socially to others and one-third do not experience these challenges (Ricketts et al., 2022).

These cases highlight some ways in which communication needs could be overlooked in children with C-TS. For both Cam and Kent, there were significant delays in the identification of their language needs and, in Kent's case, the case history shows that these indicators were not subtle in the early years (significant delay in acquiring early language milestones). A focus on providing behavioural support in both children may have meant that aspects of communication development that could have been strengthened with early intervention were overlooked. There is considerable literature confirming that children with emotional and behaviour issues often have unidentified language needs (Cole, 2001; Hollo et al., 2013; Korrel et al., 2017; Toppelberg & Shapiro, 2000). It would be important for health services to facilitate a thorough investigation

into language development when families are accessing services for emotional and behavioural needs. In particular, children with childhood language disorders and C-TS have a complex presentation; a focus on symptoms that are more obvious and disruptive to others may put them at high risk of having their language needs under-identified and under-treated.

For Lorenzo, many excellent communication skills were evident from test scores and based on his mother's descriptions of his communication across contexts. His mother also noted unique social communication characteristics and Lorenzo's test scores on the CCC-2 seemed to reflect these features of this communication (i.e., initiation, scripted language, context, and interests). Lorenzo's mother indicated that his social communication style was creating strain in his relationships with adults (especially at school). Parents, teachers, and health professionals may not be aware that the SLP can provide support when children are experiencing challenges specific to social communication. Advocacy and education about the SLP's role in supporting social communication development is an important area that applies to supporting children with C-TS as well as other groups of individuals. Further, and in line with the intuition of parents in this study, children's social differences must be accepted rather than pathologized as SLPs provide intervention for social communication development. This approach has been termed 'neurodiversity-affirming' and is being promoted in strengths-based Autism literature (e.g., Gaddy & Crow, 2023; Oates & Bean, 2023).

In Jasper's case, his mother described difficulties expressing himself (e.g., being unable to explain why he was refusing to stay at school one day). Jasper's test scores reflected strong expressive language and strengths in several other areas, but also reflected challenges specific to Recalling Sentences, and social-pragmatic language, and revealed some discrepancy between Narrative Production and Narrative Comprehension (with comprehension being average and

production being low average). Jasper's case demonstrates inconsistency between language use in day-to-day situations and test scores and underscores the importance of considering day-to-day communication when evaluating communication.

Based on these five cases, we can conclude that these standardized assessments would be appropriate for clinical use when children with C-TS are referred to the SLP. The CELF-5 Core Language Score was effective in identifying Kent's language disorder (but it did not identify the communication challenges of other participants). The TNL-2 and CASL-2 Supralinguistic Index were both effective in identifying the children with language disorders (Kent and Cam). The CCC-2 General Communication Composite effectively identified language disorders (Kent and Cam) and children who had needs in a specific area of communication (Jasper and Lorenzo). The disadvantage of the CCC-2 was that the General Communication Composite did not provide information about the very strong communication skills that Jasper and Lorenzo had in certain areas of their development. In clinical practice, using a combination of these tools augmented with information about day-to-day communication would provide a comprehensive clinical assessment for a child with C-TS.

Having an SLP as a regular member of the care team for children with C-TS would be beneficial to provide ongoing support for children with co-occurring language disorders and to address other communication needs that come up (e.g., periods of mutism, support with social communication development, literacy learning challenges). It is important to note that TS is often not diagnosed until middle childhood (tics are usually seen around 5-6 years and a diagnosis of TS is given around 8 years). Ideally, communication needs would be identified long before TS is diagnosed, thus paving the way for intervention to be provided. Further, the insights and experiences of parents suggest that SLPs will need to take a flexible approach,

acknowledging that TS and related symptoms can vary day-to-day and that they can get in the way of participation and practice in traditional models of therapy.

Limitations

Although this project allowed for an in-depth investigation of specific examples of communication development in C-TS, the particular experiences of these participants may not generalize to larger groups of children with C-TS or to individuals with characteristics that differ from these cases.

Information was collected mostly from parents in this study and therefore the findings do not represent the perspectives of a broad range of people (e.g., children themselves, peers, teachers). It is important to note that parents and children often perceive the challenges associated with TS differently. For example, prior research has indicated that parents perceived rage attacks and motor tics as more bothersome and in need of intervention compared to children (Ghanizadeh et al., 2010). Health-related quality of life may be perceived differently through the eyes of parents and children with TS as well (Cavanna et al., 2013; Gutierrez-Colina et al., 2015).

Future Research Directions

Future research should aim to gain a greater understanding of how to tailor treatment strategies to meet the needs of children with C-TS. More research is needed to develop practice guidelines for SLPs supporting children with C-TS in clinical settings. With more research, such guidelines can be developed. Larger studies measuring specific areas of language development will be helpful to make generalizations about communication in children with TS. Implementation science approaches can be used to ensure that SLPs have the knowledge they

need to assess and support language and communication development and to ensure that health services are set up to offer children early supports when they would be of benefit.

Future research should aim to understand the first-hand communication experiences of individuals with TS. Incorporating knowledge about lived experience will be important for establishing priorities going forward. Future work focused on social behaviour and social functioning should aim to understand masking behaviours for this group of individuals, considering its purpose and potential impacts on mental health.

Conclusion

This study has highlighted many barriers and facilitators to language and communication development and how they manifest the day-to-day contexts of a variety of cases of C-TS. The complexity and variability across children and contexts in this study demonstrate the importance of tailoring assessment and support services to the individual child.

References

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). https://doi.org/10.1176/appi.books.9780890425596

Bates, E. (1991). A Functionalist Approach to Language and its Implications for Assessment and Intervention, In T. Gallaher (Ed). *Pragmatics of Language: Clinical Practice Issues*. Springer.

Bishop, D. (2003). *Children's Communication Checklist (CCC)* (2nd ed.). Pearson.

Braun, V., & Clarke, V. (2021). Can I use TA? Should I use TA? Should I not use TA? Comparing reflexive thematic analysis and other pattern-based qualitative analytic approaches. *Counselling and Psychotherapy Research, 21*(1), 37–47. https://doi.org/10.1002/capr.12360

Braun, V., & Clarke, V. (2022). *Thematic analysis: A practical guide*. Sage.

Bronfenbrenner, U. (1979). *The ecology of human development: Experiments by nature and design*. Harvard University Press.

Carrow-Woolfolk, E. (2017). *The Comprehensive Assessment of Spoken Language (CASL-2)* (2nd ed.). WPS.

Carter, A. S., O'Donnell, D. A., Scahill, L., Schultz, R. T., Leckman, J. F. & Pauls, D. L. (2000). Social and emotional adjustment in children affected with Gilles de la Tourette's Syndrome: Associations with ADHD and family functioning. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 41*(2), 215–223. https://doi.org/10.1017/S0021963099005156

Cath, D. C., Muller-Vahl, K. R., & Mathews, C. A. (2022). Other psychiatric comorbidities in Tourette syndrome. In D. Martino & J. F. Leckman (Eds.), *Tourette syndrome* (2nd ed., pp. 65–94). Oxford University Press.

Cavanna, A. E., Luoni, C., Selvini, C., Blangiardo, R., Eddy, C. M., Silvestri, P. R., Cali', P. V., Gagliardi, E., Balottin, U., Cardona, F., Rizzo, R., & Termine, C. (2013). Parent and self-report health-related quality of life measures in young patients with Tourette syndrome. *Journal of Child Neurology, 28*(10), 1305–1308. https://doi.org/10.1177/0883073812457462

Channon, S., Pratt, P., & Robertson, M. M. (2003). Executive function, memory, and learning in Tourette's syndrome. *Neuropsychology, 17*(2), 247–254. https://doi.org/10.1037/0894-4105.17.2.247

Cole, D. (2001). Narrative development in aggressive boys. *Behavioral Disorders, 26*(4), 332–343. https://doi.org/10.1177/019874290102600401

Constantino, J. N., & Gruber, C. P. (2012). *Social Responsiveness Scale (SRS)* (2nd ed.). Western Psychological Services.

Cooper, C., Robertson, M. M., & Livingston, G. (2003). Psychological morbidity and caregiver burden in parents of children with Tourette's disorder and psychiatric comorbidity. *Journal of the American Academy of Child & Adolescent Psychiatry, 42*, 1370– 1375. https://doi.org/10.1097/01.CHI.0000085751.71002.48

Cravedi, E., Deniau, E., Giannitelli, M., Pellerin, H., Czernecki, V., Priou, T., Xavier, J., Consoli, A., Hartmann, A. & Cohen, D. (2018). Disentangling Tourette syndrome heterogeneity through hierarchical ascendant clustering. *Developmental Medicine and Child Neurology, 60*(9), 942–950. https://doi.org/10.1111/dmcn.13 913

Creswell, J. W., & Plano Clark, V. (2018). *Designing and conducting mixed methods research* (3rd ed.). Sage.

Crotty, M. (1998). *The foundations of social research: Meaning and perspectives in the research process*. Sage.

De Groot, C. M., Yeates, K. O., Baker, G. B., & Bornstein, R. A. (1997). Impaired neuropsychological functioning in Tourette's syndrome subjects with co-occurring obsessive-compulsive and attention deficit symptoms. *Journal of Neuropsychiatry and Clinical Neurosciences, 9*(2), 267–272. https://doi.org/10.1176/jnp.9.2.267

Drury, H., Channon, S., Barrett, R., Young, M. B., Stern, J. S., Simmons, H., & Crawford, S. (2012). Emotional processing and executive functioning in children and adults with Tourette's syndrome. *Child Neuropsychology, 18*(3), 281–298. https://doi.org/10.1080/09297049.2011.613811.

Eapen, V., McPherson, S., Karlov, L., Nicholls, L., Črnčec, R., & Mulligan, A. (2019). Social communication deficits and restricted repetitive behavior symptoms in Tourette syndrome. *Neuropsychiatric Disease and Treatment, 15*, 2151–2160. https://doi.org/10.2147/ndt.s210227

Feehan, A., & Charest, M. (2023). A scoping review of oral language and social communication abilities in children with Tourette syndrome. *International Journal of Language and Communication Disorders, 59*(1), 1–22. https://doi.org/10.1111/1460-6984.12949

Fetters M. D. (2020). *The mixed methods research workbook: Activities for designing, implementing, and publishing studies*. Sage.

Fetters, M. D. (2022). A comprehensive taxonomy of research designs, a scaffolded design figure for depicting essential dimensions, and recommendations for achieving design naming

conventions in the field of mixed methods research. *Journal of Mixed Methods Research, 16*(4), 394-411. https://doi.org/10.1177/15586898221131238

Fitcher, B. (2014). *Homeschooled with Tourette's.* https://mollygreen.com/blog/homeschooled-with-tourettes/

Gaddy, C., & Crow, H. (2023). A primer on neurodiversity-affirming speech and language services for autistic individuals. *Perspectives of the ASHA Special Interest Groups, 8*(6), 1220-1237. https://doi.org/10.1044/2023_persp-23-00106

Gadow, K.D. & Paolicelli, L. M. (1986). *The Global Tic Rating Scale (GTRS).* State University of New York Department of Psychiatry.

Ghanizadeh, A., Mohammadi, M. R., & Dehbozorgi, G. R. (2010). Children and their parent's perceptions of symptom severity and treatment preference for Tourette syndrome. *Iranian Journal of Psychiatry, 5*(3), 93–936.

Gillam, R. B., & Pearson, N. A. (2017). *Test of Narrative Language (TNL)* (2nd ed.). WPS.

Goodman, R. (1997). *Strengths and Difficulties Questionnaire (SDQ)* [Database record].

Goodman, R., Renfrew, D., & Mullick, M. (2000). Predicting type of psychiatric disorder from Strengths and Difficulties Questionnaire (SDQ) scores in child mental health clinics in London and Dhaka. *European Child and Adolescent Psychiatry, 9*, 129–134.

Goodman, R., Ford, T., Corbin, T., & Meltzer, H. (2004). Using the Strengths and Difficulties Questionnaire (SDQ) multi-informant algorithm to screen looked-after children for psychiatric disorders. *European Child and Adolescent Psychiatry, 13*, 25–31.

Güler, A. S., Berkem, M., Yazgan, Y., & Kalaça, S. (2015). Cognitive flexibility and social responsiveness in children and adolescents with Tourette syndrome. *Child Psychiatry and Human Development, 46*(6), 940–950. https://doi.org/10.1007/s10578-015-0533-3

Gutierrez-Colina, A. M., LaMotte, J., Eaton, C., Kardon, P., & Blount, R. L. (2015). The role of self-competence in health-related quality of life and behavioral functioning of children with Tourette syndrome. *Journal of Developmental and Behavioral Pediatrics, 36*(9), 743–751. https://doi.org/10.1097/DBP.0000000000000214

Hollo, A., Wehby, J. H., & Oliver, R. M. (2013). Unidentified language deficits in children with emotional and behavioral disorders: A meta-analysis. *Exceptional Children, 80*(2), 169–186. https://doi.org/10.1177/001440291408000203

Houghton, C., Murphy, K., Shaw, D., Casey, D. (2015). Qualitative case study data analysis: An example from practice. *Nurse Researcher, 22*(5), 8-12. doi: 10.7748/nr.22.5.8.e1307. PMID: 25976531.

Hulbert, T. (1986). *Neuropsychological subgrouping of Tourette children* [Unpublished doctoral dissertation]. Wayne State University. https://doi.org/10.1017/CBO9781107415324.004

Hyter, Y. (2017). Pragmatic assessment and intervention in children. In L. Cummins (Ed.), *Research in Clinical Pragmatics* (pp. 493-526). Springer International Publishing.

Jeans, A. D. (2016). *The school and social experiences of Tourette syndrome through the first-person perspective of youth: A theatre-based approach to community-based research* [Master's Thesis]. University of Alberta.

Jensen, J. (2004). Neuropsychological and behavioural characteristics of children and adolescents with Tourette Syndrome compared to controls with dyslexia. *Journal of Allergy and Clinical Immunology, 130*(2), 556. https://doi.org/10.1016/j.jaci.2012.05.050

Korrel, H., Mueller, K. L., Silk, T., Anderson, V., & Sciberras, E. (2017). Research review: Language problems in children with Attention-Deficit Hyperactivity Disorder – a

systematic meta-analytic review. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 58*(6), 640–654. https://doi.org/10.1111/jcpp.12688

Lee, M. Y., Mu, P. F., Wang, W. S., & Wang, H. S. (2016). “Living with tics”: Self-experience of adolescents with Tourette syndrome during peer interaction. *Journal of Clinical Nursing, 25*(3–4), 463–471. https://doi.org/10.1111/jocn.13074.

Legg, C., Penn, C., Temlett, J., & Sonnenberg, B. (2005). Language skills of adolescents with Tourette Syndrome. *Clinical Linguistics and Phonetics, 19*(1), 15–33. https://doi.org/10.1080/02699200410001665270

Lincoln, Y. S., & Guba E. G. (1985). *Naturalistic Inquiry*. SAGE.

Ludlow, A. K., Cutler, A., & Keville, S. (2022). Teachers' knowledge and awareness of Tourette syndrome in children attending mainstream school. *Journal of Research in Special Educational Needs, 22*(4), 323-331.

Miles, M. B. & Huberman, A. M. (1994). *Qualitative data analysis: An expanded sourcebook* (2nd ed.). Sage.

Morse, J. M. (1994). ‘Emerging from the data’: the cognitive processes of analysis in qualitative inquiry. In J. M. Morse (Ed.), *Critical issues in qualitative research methods.* Sage.

Morse, J. M., Barrett, M., Mayan, M., Olson, K., & Spiers, J. (2002). Verification Strategies for Establishing Reliability and Validity in Qualitative Research. *International Journal of Qualitative Methods, 1*(2), 13–22. https://doi.org/10.1177/160940690200100202

Murphy, T. & Eddy, C. M. (2013). Neuropsychological assessment in Tourette syndrome. In: D. Martino & J. F. Leckman (Eds.), *Tourette syndrome*. Oxford University Press, pp. 439–467.

Nussey, C., Pistrang, N., & Murphy, T. (2014). Does it help to talk about tics? An evaluation of a classroom presentation about Tourette syndrome. *Child and Adolescent Mental Health, 19*(1), 31–38. https://doi.org/10.1111/camh.12000

O'Quinn, A. N., & Thompson, R. J. (1980). Tourette's syndrome: An expanded view. *Pediatrics, 66*(3), 420–424. https://doi.org/10.1016/s0002-7138(09)61008-6

Oates, M., & Bean, A. (2023). Qualitative Analysis of the Experiences and Perspectives of Autistic Speech-Language Pathologists. *American Journal of Speech-Language Pathology, 32*(5), 2178-2191. https://doi.org/10.1044/2023_AJSLP-22-00399

Pine, R., Bruckner, K., Mbinta, J., Twemlow, R., & Anderson, S. (2022). Perceptions of parents and caregivers in New Zealand: Educational experiences of their children with Tourette syndrome. *International Journal of Disability, Development and Education, 71*(3), 1–14. https://doi.org/10.1080/1034912X.2022.2120603

Richardson, L. (2000). Writing: A method of inquiry. In N. K. Denzin & Y. S. Lincoln (Eds.), *Handbook of qualitative research* (2nd ed., pp. 923-948). Sage.

Ricketts, E. J., Wolicki, S. B., Danielson, M. L., Rozenman, M., McGuire, J. F., Piacentini, J., Mink, J. W., Walkup, J. T., Woods, D. W., Bitsko, R. H. (2022). Academic, interpersonal, recreational, and family impairment in children with Tourette syndrome and Attention-Deficit/Hyperactivity Disorder. *Child Psychiatry and Human Development, 53*(1), 3–15. https://doi.org/10.1007/s10578-020-01111-4

Robinson, L., Bitsko, R., Schieve, L., & Visser, R. (2013). Tourette syndrome, parenting aggravation, and the contribution of co-occurring conditions among a nationally representative sample. *Disability and Health Journal, 6*(1), 26–35. https://doi.org/10.1016/j.dhjo.2012.10.002

Rothenberger, A., & Roessner, V. (2013). The phenomenology of Attention-Deficit/Hyperactivity Disorder in Tourette syndrome. In D. Martino & J. F. Leckman (Eds.), *Tourette Syndrome* (pp. 439-467). Oxford University Press.

Rutter, M., Bailey, A., & Lord, C. (2003). *The Social Communication Questionnaire (SCQ).* Western Psychological Services.

Schuerholz, L. J., Singer, H. S. & Denckla, M. B. (1998). Gender study of neuropsychological and neuromotor function in children with Tourette syndrome with and without attention-deficit hyperactivity disorder. *Journal of Child Neurology, 13*(6), 277–282. https://doi.org/10.1177/088307389801300607

Spencer, T., Biederman, J., Harding, M., O'Donnell, D., Wilens, T., Faraone, S., Coffey, B., & Geller, D. (1998). Disentangling the overlap between Tourette's disorder and ADHD. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 39*(7), 1037–1044. https://doi.org/10.1017/S0021963098002984

Stake, R. E. (2006). *Multiple Case Study Analysis.* The Guilford Press.

Stake, R. E. (1995). *The Art of Case Study Analysis*. Sage.

Stewart, S., Greene, D., Lessov-Schlaggar, C., Church, J., & Schlaggar, B. (2015). Clinical correlates of parenting stress in children with Tourette syndrome and in typically developing children. *The Journal of Pediatrics, 166*(5), 1297–1302e3. https://doi.org/10.1016/j.jpeds.2015.01.041

Suh, H. W., Yoon, S. I., Hong, S., Lee, H. W., Lee, M., Kim, J. W., & Chung, S. Y. (2022). How do children with Tourette's syndrome and their caregivers live with the disorder? A systematic review of qualitative evidence. *Frontiers in Psychiatry, 13*(Sep), 1–16. https://doi.org/10.3389/fpsyt.2022.992905

Sukhodolsky, D. G., Scahill, L., Zhang, H., Peterson, B. S., King, R. A., Lombroso, P. J., Katsovich, L., Findley, D. & Leckman, J. F. (2003). Disruptive behavior in children with Tourette's syndrome: Association with ADHD comorbidity, tic severity, and functional impairment. *Journal of the American Academy of Child and Adolescent Psychiatry, 42*(1), 98–105. https://doi.org/10.1097/00004583- 200301000-00016

Thompson, R. J., O'Quinn, A. N., & Logue, P. E. (1979). Gilles de la Tourette syndrome: A review and neuropsychological aspects of four cases. *Journal of Pediatric Psychology, 74*(4), 371-387. https://doi.org/10.1177/014107688607900508

Thorne, S. (2016). *Interpretive Description*. Left Coast Press.

Toppelberg, C. O., & Shapiro, T. (2000). Language disorders: A 10-year research update review. *Journal of the American Academy of Child and Adolescent Psychiatry, 39*(2), 143–152. https://doi.org/10.1097/00004583-200002000-00011

Tourette Association of America. (2021). *Homeschooling: A flexible, fluid, child-focused education*. https://tourette.org/homeschool/

Travis, E., & Juarez-Paz, A. V. O. (2020). Experiences of Tourette syndrome caregivers with supportive communication. *Qualitative Health Research, 30*(10), 1480–1490. https://doi.org/10.1177/1049732320915444

Wadman, R., Glazebrook, C., Beer, C., & Jackson, G. M. (2016). Difficulties experienced by young people with Tourette syndrome in secondary school: A mixed methods description of self, parent and staff perspectives. *BMC Psychiatry, 16*(1), 1–11. https://doi.org/10.1186/s12888-016-0717-9

Wiig, E. H., Semel, E., & Secord, W. A. (2013). *Clinical Evaluation of Language Fundamentals (CELF-5)* (5th ed.). Pearson.

Wiig, E. H., Semel, E., & Secord, W. A. (2013). *Clinical Evaluation of Language Fundamentals Screening Test* (5th ed.). Pearson.

Chapter 6: Methods Supplement for Communication in Children with Complex Tourette Syndrome: Insights from Five Mixed Methods Case Studies (Study 3)

This chapter provides additional detail about the methods used in Study 3. Mixed methods projects require a careful integration of quantitative and qualitative data (Schoonenboom & Johnson, 2017). The two types of data complemented each other and mixing occurred throughout the data collection stage and during analysis. The complementarity between the two types of data occurred because objective knowledge ("looking at") and subjective knowledge ("looking in") were combined, thereby aiming to obtain a more holistic view (Carroll & Rothe, 2010).

The bioecological model was considered throughout Study 3, with a focus on the "individual" and the "microsystem" contexts. Individual factors were investigated through the collection of background information about the child's diagnosis; cognitive factors were measured through language testing and the collection of intelligence and past language testing results; and day-to-day communication interaction factors were investigated through the parent log book and the parent interview. Home and community contextual factors of the microsystem were investigated from the parents' perspectives through interview questions about home interactions, community life, and school. Furthermore, the constructionist framework used in the qualitative portion of the project allowed for relationships between factors to be brought to light and while preserving the complexity of specific examples.

Selecting Quantitative and Qualitative Data Collection Tools

Quantitative Measure of Parent-Observed Communication

The Children's Communication Checklist, Second Edition (CCC-2; was selected as a measure of parent-observed communication because it provides a total communication composite score, a social-pragmatic composite score, as well as measures for skills in specific domains of communication that are of interest for children with TS (see Table 8 for a summary of the psychometric properties of the CCC-2)

Quantitative Measure of Child Language Skills

The Clinical Evaluation of Language Fundamentals, Fifth Edition (CELF-5; Wiig et al., 2013) Core language subtests (30-45 mins) were selected as a measure of language skills. The subtests include Word Classes (the child must choose two words that are related to one another from a group of three or four), Formulated Sentences (the child is asked to formulate a sentence about a picture using a target word given by the examiner), Recalling Sentences (the child repeats a sentence that has been said aloud by the examiner), and Understanding Spoken Paragraphs (The child answers questions about paragraphs read aloud to them). Children completed three of these four subtests to receive a Core Language Index score (subtests varying with age). The CELF-5 provides detailed administration and scoring instructions. Normative data were collected with a sample of 3000 5–21 year-old individuals, representative of the US population. Internal consistency index coefficients for the core language subtests range from .77-.95. Core language Index coefficients range from .94-.97. Test-retest reliability *r* for core language subtests ranged from .69-.90 and Core language index *r* ranged from .88-.92. Inter-rater reliabilities ranged from .91-.99. Content validity is demonstrated by presenting a literature review, obtaining user feedback, and completing an expert review. The Core Language Score of

the CELF-5 demonstrates the ability to differentiate between children with language disorders and children with typical language development, showing a large effect size. The optimal cut score, representing a sensitivity and specificity of .97, is 1.3 SD below the mean (a score of 80).

Quantitative Measure of Narrative Language

The Test of Narrative Language, Second Edition (TNL-2; Gillam & Pearson, 2017; 15-20 mins) was selected as a test of narrative language development. The TNL-2 provides detailed administration and scoring instructions. The TNL-2 was standardized on a sample of 1,310 children representative of the US population. Internal consistency reliability coefficients ranged from .73 and .91 for subtest scores and .85 and .94 for index scores. Test-retest reliability correlations were .73-.93 for Narrative Language Ability Index. Scorer difference reliability coefficients for interrater reliability were above .90 for subtests and index scoring. A cut-off score of 92 provides a sensitivity of .92 and a specificity of .92 for detecting Developmental Language Disorder.

Quantitative Measure of High-Level Language

The Metalinguistics subtests of the Comprehensive Assessment of Spoken Language (CASL-2; Carrow-Woolfolk, 2017; 40 mins) were selected as a measure of high-level language. The CASL-2 provides detailed administration and scoring instructions. It was standardized on a sample of 2,394 people representative of the US population excluding gifted or severely disabled people. Internal consistency reliability coefficients were between .85 and .99 for subtest scores and .88 and .96 for index scores. Test-retest reliability correlations were .73-.94 for test scores and .88-.96 for index scores. Intraclass correlation coefficients for interrater reliability were .86-.97. Convergent reliability was demonstrated through correlating the results of the CASL-2 with other clinical assessments. A cut-off of 85 on the Supralinguistic Index represents a sensitivity of

.74 and specificity of .84. MANOVA was used to compare means of test and index scores for children from the normative sample with children who have expressive/receptive language disorder, hearing impairment, ASD, social-pragmatic communication disorder, intellectual disability, learning disorder, and developmental delay. Scores were significantly lower across all comparisons. Effect sizes were all large. Means were 76 (ASD), 75 (SPCD), and 82 (LD).

Qualitative Parent Log Book About Day-to-Day Child Communication

A 7-day log book was selected to gather qualitative examples of the child's day-to-day communication. Parents were asked to fill out the log book because they know the children well and observe them regularly. A daily log book requires parents to recount only recent events and may lead to high accuracy and detail in reporting (Parke et al., 2019). Parents completed a 7-day log book with daily examples of their child's communication challenges and successes. They were asked 1) to provide an example of instances when the child communicated their message successfully; 2) to provide an example of when the child had difficulties communicating; 3) to make note of what else was going on in each situation; and 4) to note the reactions or responses of others who were present. See an example of this form in Appendix D.

Qualitative Semi-Structured Parent Interviews About Child Communication

Qualitative semi-structured interviews were selected to collect information about child communication in different contexts and barriers and facilitators to communication. A qualitative interview is a conversation between a researcher and an informant/participant. The researcher attempts to understand the subjective reality of the interviewee on the phenomenon of interest to answer research questions (Creswell & Poth, 2018). A semi-structured format provides some structure to guide respondents and encourages elaborating on examples but uses open-ended questions so that participants have space to discuss what they believe is relevant to the topic.

Using an initial interview guide (see Appendix E), log book examples and language testing results were used to develop an individualized interview guide for each participant. Interviews lasted 60 minutes and were conducted in a video meeting. All interviews were recorded using a Marantz audio recorder.

Rigor Practices

To produce high-qualitative research, the use of specific rigor practices is necessary. Morse et al. (2002) provide a list of strategies that can be used throughout the research process, including investigator responsiveness, methodological coherence, appropriate sampling, and an active analytic stance.

Investigator Responsiveness

Detailed notes about the data collection plan, any changes being made, and the reason for these decisions were kept in an audit trail document. For example, a need for more background information was identified during data collection with the first two participants, and therefore, the background questionnaire was expanded at this stage in the project. Additional records were also requested at this time, including the results of intellectual testing, previous speech-language pathology testing scores/dates, and dates and details of previous intervention periods with an SLP. Through the creation of reflexive memos noting initial thoughts and impressions about findings, it was possible to move beyond initial impressions to gain a deeper understanding of aspects of the cases and what they could teach us. As an example, I had initially noted that parents found it relevant to mention that their child was able to suppress tics when in public. This seemed to indicate that suppression of tics in public may be an important feature of communication in children with TS in some environments of the micro-system. However, moving beyond this surface-level analysis and deeper into this aspect of parent interviews

allowed me to interpret this piece of information in connection with the child's public identity, saving face with the outside world, and the importance of the child having a place and people with whom they could be their authentic selves (somewhere to let their tics out).

Another challenge that required responsiveness was the process of carrying the bioecological model through each aspect of the project – from planning data collection techniques, to interview planning, to coding transcripts and analysis – while also leaving space for bottom-up developments and interpretations. When coding, it was important to temporarily set aside the idea that each piece of transcript should fit somewhere within the model, because this would have threatened the depth of the analysis. Therefore, a dash with words like 'home' 'school' and 'peers' was added to codes when needed, but full flexibility and creativity were allowed in the wording of the codes themselves.

Methodological Coherence

Data collection methods were carefully chosen to provide insight into the research question, which centered around collecting information about the child's day-to-day communication in their micro-system (school, home, and peers). Aspects of this have been described throughout the introductory chapter of this thesis. By being responsive and making additions to information that was being collected throughout the project where needed, the project maintained coherence.

Appropriate Sampling

Parent-child dyads were chosen purposefully for this research. Since children with complex TS (those with co-occurring conditions) are more likely to have challenges in language development, these were the children of interest. The purpose was not to isolate the contribution of TS and other conditions to the child's communication challenges but to get a holistic picture

of communication functioning for specific cases when multiple complex factors are at play. A variety of cases were selected to present a range of presentations of communication in complex TS. Two of the children were dealing with persistent language disorders, two had recovered from language difficulties, and one had never been identified as having any language problems. Five cases were thought to be sufficient to provide a variety of in-depth case descriptions, but not so many as to threaten the researcher's ability to provide depth in individual cases. In keeping with the assumption inherent to interpretive description, namely that individual experiences of a phenomenon can vary infinitely, saturation was not a goal (Thompson Burdline et al., 2021) .

Active Analytic Stance

The analytic work of the researcher fits into all of the topics already discussed: responsiveness throughout the project, the maintenance of methodological coherence, and sampling methods. Maintaining a reflexivity journal throughout the project facilitated continual efforts at being reflexive about my positionality and how it was shaping my interpretations was an important component of maintaining an active analytic stance. Finally, integrating theory into the research process and considering how the results can build on current theory shaped the analysis.

Chapter 7: General Discussion

This thesis addressed aspects of communication in children with TS using three studies. The first study was a scoping review of language and social communication in children with TS (Chapter 2). The second study gathered parent-reported information about communication and psychosocial functioning (Chapter 3). The third study was a series of mixed methods case studies that aimed to describe the day-to-day communication of five children with Complex TS (Chapter 5). In this general discussion chapter, I will begin with a summary of each study and then address the combined findings of this thesis with regard to the communication profiles of children with TS. Finally, I will discuss the implications for clinical practice for speech-language pathologists (SLPs), future research directions, and limitations of this research.

Summary of the Research

Chapter 2 mapped the scope and landscape of existing literature on language and social communication skills in children with TS using a scoping review. Through a review of the 56 existing studies that examined the skills of children with TS, we concluded that several areas of language development appear to be strong in children with TS. There were nonetheless clues as to where language development challenges might manifest and existing studies pointed toward social communication difficulties. The scoping study also identified that further research is needed in order to understand language and social communication development in children with TS and should focus on gathering information about domains (i.e., form, use, content) and modalities (i.e., expressive and receptive) of language development and observation of social communication and social cognition skills. The findings of the scoping study were used to inform the two subsequent studies.

In Chapter 3 we described a study in which we measured parent-reported communication and psychosocial skills in a sample of 61 children, the majority of whom had co-occurring conditions. I measured the proportion of children who had parent-reported communication scores below the average range and whether or not communication scores differed from the scores of a normative sample. I also addressed whether or not co-occurring ADHD, OCD, and/or anxiety contributed to lower communication scores in this sample. Finally, I examined the relationship between levels of parent-reported psychosocial functioning and communication functioning for children with low, medium, and high tic disorder severity. I found that 38% of children with TS had communication challenges and that the group as a whole had lower communication scores compared to published normative data. No single co-occurring condition contributed significantly to communication challenges; however, lower communication scores were related to higher psychosocial difficulties. Children with low tic disorder severity tended to have higher communication skills and lower levels of psychosocial challenge, whereas children with high tic disorder severity tended to have lower communication skills and higher levels of psychosocial challenge.

Chapter 5 presents a study of communication in five children with TS using standardized testing and parent-reported information to explore how communication is facilitated or impeded in day-to-day functional contexts. I used the bioecological model to understand how child characteristics interact with contextual environments (microsystem: school, home, and peers) to produce successful or unsuccessful communicative interactions. A focus was placed on functionalism, or how the child's communication plays out in real-life contexts. The results highlighted the importance of educational accommodations or alternative learning environments, the need for parent management of interactions in the home including fostering a calm and

accepting atmosphere, and, in some cases, the need for support and/or coaching with social interactions.

Bringing it all together: Summarizing communication profiles in children with TS

The case studies in this thesis illustrated that a wide variety of communication profiles are possible in children with TS. Children can experience variability with respect to concerns relevant to speech-language pathology practice, such as challenges in reading development, social interaction, periods of mutism, and language development challenges. Given this diversity, what generalizations can be made about communication in children with TS? Our findings supported past conclusions, strengthening the evidence that social communication differences are common in this group and that approximately one-third of children with complex TS may have language delays/disorders (Darrow et al., 2017; Cravedi et al., 2018; Güler et al., 2015; Spencer et al., 1998). Our findings also paralleled results from past studies about potential challenges in high-level language and language coherence; however, lower mean scores were observed in nearly all of the communication domains tested by the Children's Communication Checklist, Second Edition (CCC-2; Bishop, 2003), suggesting that difficulties, for those who have them, may not be limited to those areas. Finally, the fact that many children received scores within the mean range on the individual domains of the CCC-2 (at least half of the sample for each domain) supports the conclusions from the scoping study that children with TS have many strengths in their communication development. Syntax was identified as a strength in the parent-reported measures study and verbal fluency, verbal IQ, vocabulary, categorization, and ability to recall/repeat language were strengths identified in the scoping study.

Creating a definitive communication profile for children with TS as a group is likely not possible given the variation that appears to exist across children. However, we can conclude that communication assessments should be offered to children with complex TS because we can

assume that many of these children will go on to have potential communication goals that SLPs can support.

Clinical Implications

Clinical implications will be discussed by addressing four key areas: addressing the tendency toward underidentification of communication challenges in children with TS, assessment considerations, treatment considerations, and anti-ablism in speech-language pathology practice with children with TS.

Avoiding Underidentification

In study 3, parent-reported data and case studies both pointed to communication delays and difficulties that had gone unidentified. Communication challenges in early childhood have been connected with later problems in behavioral adjustment (Bornstein et al., 2013; Yew & O'Kearney, 2013). Similarly, problems specific to social communication in early childhood have been connected to a range of later emotional and behavioural challenges (Dall et al., 2022). These associations underscore the importance of early identification of communication challenges and skill-building interventions. Strategies for early identification; however, need to consider that tics tend to emerge in middle childhood (around 7-8 years) and most children are diagnosed with TS after the age of eight (Leckman et al., 2022). If communication needs are identified after TS diagnosis, children may have missed out on opportunities to receive beneficial intervention at earlier periods in childhood. Thus, communication assessment and intervention should also be available into the school years so that children whose communication needs were not detected at earlier ages can be identified and offered appropriate support.

For pediatric SLPs offering early assessment and treatment, it is important to note that attentional difficulties are often the sole early childhood symptom shown by children with TS (at

age 2-3; Leckman et al., 2022). As it the case for childhood language disorders generally, family and perinatal history can offer important information regarding risk factors (Paul et al., 2017). Children are at higher risk for TS when they have a family member with TS, if there were complications during pregnancy, delivery or the neonatal period, and if they had a low birth weight (Abdallah & Fernandez, 2022; Hoekstra & Dietrich, 2022). In addition to providing supports and interventions for communication, SLPs can also play a role in educating other health and education professionals about how TS can present.

Assessment Considerations

For children with TS, case history information must include information about co-occurring conditions. Clinicians should aim to gather a thorough list of conditions to understand the child's profile. This will support goal selection and intervention planning. For example, the SLP may assess executive functioning skills for children with co-occurring ADHD (Roitsch, 2019). For children with social anxiety, social language may be an area to focus on (Halls et al., 2015).

Children with TS show a range of strengths and some possible areas of challenge in communication development. In assessment, the SLP should look beyond language tasks that assess vocabulary knowledge and performance on single-word tasks and consider narrative language, coherence, high-level language, and social communication. When the SLP is considering standardized tools for the assessment of communication in children with TS, a few specific recommendations can be made. The CELF-5 Core Language subtests did not consistently capture communication challenges in case study participants who had reported and/or confirmed communication needs. For one participant, both TNL-2 and CASL-2 Supralinguistic scores were more consistent with parent-reported communication development,

language disorder diagnosis, and the level of support the child received for language development. It would be beneficial in clinical practice to include instruments that assess narrative language and high-level language skills (e.g., inferencing and non-literal language) such as the TNL-2 and CASL-2. These can supplement core language testing and increase the likelihood of capturing difficulties that may be missed if one does not look beyond semantics or structural language. For both of these case study participants, the CCC-2 score reflected communication challenges. One participant had excellent language skills in most areas but was experiencing difficulties fin social communication. For him, the CCC-2 was the only assessment instrument that captured areas of need. Furthermore, in Study 2, the CCC-2 parent questionnaire suggested identification rates similar to those previously reported for children with complex TS (Cravedi et al., 2018; Spencer et al., 1998). The CCC-2 appears to be a useful tool and could be part of an intentional and comprehensive communication assessment protocol for children with TS. We used a communication log book in the mixed methods study to identify examples of both successes and difficulties in day-to-day communication. This tool could be used as a clinical assessment technique and could provide valuable insights for intervention planning.

Treatment Considerations

Parent insight demonstrated the importance of flexibility and providing a calm environment to help children maintain emotional regulation. The SLP can use this guidance as they set up the environment for interventions targeting specific communication development goals. Parent-reported measures demonstrated that children with lower communication scores also tended to have higher levels of psychosocial challenge and may experience a higher severity in their tic disorders. Clinicians can expect to encounter children with psychosocial challenges and should be prepared with proactive strategies (Katsovich et al., 2003). Some general strategies

for supporting children with emotional and behavioural challenges include creating predictability using routines/schedules, minimizing noise and distractions, thoughtfully setting up the physical space, explaining goals and their importance, corrective and positive feedback, and positive regard (Gilkey-Hirn & Park, 2012). A strong correlation between stress/anxiety levels and TS severity has been previously reported (Leckman et al., 2022). The SLP should keep in mind that decreasing anxiety may reduce the likelihood of the onset of tics during treatment sessions.

As with other populations, communication goals for children with TS should be individualized to target specific areas of communication that emerge as needs for each unique child. Because of the diversity in this population due to many factors, including the presence of co-occurring conditions, the SLP may find that goals vary greatly between children and can vary over time with the same child.

As part of an educational team, the SLP can be instrumental in bringing observations about the child's learning, communication skills, and other characteristics to the educational team for discussions about appropriate school placement and/or accommodations. As part of a medical team, the SLP can also bring observations that will support differential diagnosis of co-occurring conditions, decisions about medication, and other medical treatment decisions.

How the SLP Views TS: Anti-ableism

Individuals with TS can differ from neurotypical people in their social behaviour and social thinking. Parents of case study participants expressed the need to support their child by way of accepting the child's characteristics and ways of thinking. As SLPs, it is important to continuously ask whether or not any communication differences we observe require intervention, and, if we find that they do, why? Interventions aimed at targeting certain communication characteristics measured by the CCC-2 (such as 'interests' and 'scripted language') lend

themselves toward a focus on training the child to act more like neurotypical peers. We can learn from the Autistic community in which many individuals have expressed that such interventions have caused harm and/or trauma (e.g., Anderson, 2023). Recent understandings about how to mitigate this danger have focused on the importance of listening to and not ignoring the preferences and opinions of individuals undergoing intervention (Anderson, 2023; Cumming et al., 2020). While the SLP may choose specific intervention goals with little family input in some areas of practice (e.g., selecting a speech sound to target); it is very important to ensure that communication goal selection for children with TS involves input from parents and children.

Being anti-ableist also means taking care not to leap from detecting differences in children with TS to deficit-focused framing or to creating harmful stereotypes. For example, a tendency toward low scores on the Interests domain of the CCC-2 for children with TS could lead to a generalization that children with TS may have 'abnormal' or 'narrow' interests. Instead, we can frame these differences in positive ways. We could refer to children's interests using language like 'passions' and 'enthusiasms,' which Wilson & Prizant (2022) have used to discuss Autistic special interests. The SLP can contribute to improving societal acceptance of the differences associated with TS through language choice and ongoing discussions with clients, families, and other health and education service providers.

Limitations

Ideally, randomly selected children would have been used in the parent-reported measures study; however, a convenience sample was used due to time, access, and budget limitations. Because of this, the sample may not be representative of the population of children with TS. Similarly, the study may not have been accessible to all parents who have a child with TS. Families required computer and internet access and the time and skills to complete a series

of questionnaires. Case studies gathered a high level of detail about individual experiences; however, since they were highly specific, findings may not be generalizable to other families who have a child with TS.

Comparing children's CCC-2 scores to a normative sample in the parent-reported measures study was a cost-effective method; however, there may have been important differences between the recruited sample of children and the normative sample, which could affect the validity of comparisons. Where cost and time allow, the use of a group of nondisabled peers for comparison would be preferable.

Another limitation of these studies was that the majority of information came from parents, and therefore, findings about communication in children with TS reflect parent perspectives and not the perspectives of others such as the child themselves, friends, or teachers.

Future Research Directions

Future research on communication in children with TS should look at the perspectives of multiple stakeholders in understanding communication development (e.g., teachers, peers) and it should aim to recruit participants that are representative of families in the geographic area. Furthermore, this body of literature would benefit from the use of nondisabled participants for comparison.

Direct testing of the communication skills of children with TS is still needed to gain a greater understanding of the communication skills of children with TS, as such research is still very limited. Tools that measure expressive, receptive, narrative, and high-level language skills will provide needed insight into these aspects of communication development. Language researchers can also contribute toward furthering understandings of social cognitive skill development in children with TS through the use of tasks that tap into specific skills. A research program paralleling the work of Dr. Claire Eddy with adults with TS (e.g., Eddy et al., 2010a,

2010b, 2011, 2016, 2017; Eddy & Cavanna, 2015) using tasks modified for children could begin to improve our understanding of social cognitive development in children. Specifically, it will be important to investigate how children comprehend and interpret the intentions of others, recognize and judge social indiscretions, take the perspectives of others, and regulate emotions during stressful interpersonal interactions.

Future research investigating the relationship between tic disorder severity and communication/psychosocial function should aim to use methods that help us understand the temporal development of these different aspects. Longitudinal studies can help us to understand how communication skills, psychosocial skills, and tic disorder severity develop over time to better tease apart the complexities of this relationship and identify recommended age ranges for screening and intervention.

Current recommendations for treating communication challenges in children with TS are based on very limited evidence. Intervention research is needed to support clinicians in making evidence-informed treatment decisions. Multidisciplinary research projects will continue to be of benefit to furthering our understanding of communication development and how to support communication challenges in this population. In addition to the SLP, neuropsychology, neurology, psychology, education, and others can provide important contributions.

Knowledge Translation

The findings of Chapter 2 are now published in an academic journal and have been presented at the American Speech and Hearing Association Convention in 2023. Findings of Chapters 3 and 5 will be disseminated by publishing the results in academic journals and presenting results at North American speech-language pathology conferences. The combined findings of these three studies will be presented to faculty and students in the Faculty of Rehabilitation Medicine at the University of Alberta and in the Child and Adolescent Psychiatry

department at the Glenrose Rehabilitation Hospital. A short report summarizing the findings of all three studies will be prepared to share with Tourette Canada and the Tourette Association of America.

Conclusion

This series of three studies has increased our understanding of communication development in children with TS. Children with TS tend to have many strengths in communication; however, they are at risk for language disorders and may experience challenges in some areas of language and social language development. SLPs can play a key role in identifying communication challenges in this population and supporting communication development. Children with TS are unique to other clinical groups and are often complex; they require specific considerations. Deliberately facilitating TS acceptance and avoiding contributing to stigma and deficit focus is of particular importance. These key messages can guide SLPs in supporting children with TS and their families.

References

Abdallah, S. B., & Fernandez, T. V. (2022). Genetic susceptibility in Tourette syndrome. In D. Martino & J. F. Leckman (Eds.), *Tourette syndrome* (2nd ed., pp.1-25). Oxford University Press.

Achenbach, T. M., Becker, A., Döpfner, M., Heiervang, E., Roessner, V., Steinhausen, H. C., & Rothenberger, A. (2008). Multicultural assessment of child and adolescent psychopathology with ASEBA and SDQ instruments: Research findings, applications, and future directions. *Journal of Child Psychology and Psychiatry, and Allied Disciplines, 49*(3), 251–275. https://doi.org/10.1111/j.1469-7610.2007.01867.x

Achenbach., T. M., & Rescoral., L. A. (2001). Child Behaviour Checklist (CBCL). Springer.

Adams, C. (2005) Social communication intervention for school aged children: rationale and description. *Seminars in Speech and Language, 26*(3), 282–288. https://doi.org/10.1055/s-2005-917123

Albin, R. L. (2018). Tourette syndrome: A disorder of the social decision-making network. *Brain, 141*(2), 332-347.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). American Psychiatric Association. https://doi.org/10.1176/appi.books.9780890425596

Anderson, L. K. (2023). Autistic experiences of applied behavior analysis. *Autism, 27*(3), 737–750. https://doi.org/10.1177/13623613221118216

Arksey, H., & O'Malley, L. (2005). Scoping studies: Towards a methodological framework. International Journal of Social Research Methodology: *Theory and Practice, 8*(1), 19–32. https://doi.org/10. 1080/1364557032000119616

Aromataris, E. & Riitano, D. (2014). Constructing a search strategy and searching for evidence. *American Journal of Nursing, 114*(5), 49–56. https://doi.org/10.1097/01.NAJ.0000446779.99522.f6

Baizabal-Carvallo, J., & Jankovic, J. (2022). Sex differences in patients with Tourette syndrome. *CNS Spectrums*, 1–7. https://doi.org/10. 1017/S1092852922000074

Barnes, S., & Bloch, S. (2019). Why is measuring communication difficult? A critical review of current speech pathology concepts and measures. *Clinical Linguistics and Phonetics, 33*(3), 219-236.

Baron-Cohen, S., Jolliffe, T., Mortimore, C., & Robertson, M. (1997). Another advanced test of theory of mind: Evidence from very high functioning adults with autism or Asperger syndrome. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 38*(7), 813-822.

Baron-Cohen, S. & Robertson, M.M. (1995). Children with either autism, Gilles de la Tourette syndrome or both: Mapping cognition to specific syndromes. *Neurocase, 1*(2), 101–104. https://doi.org/10. 1080/13554799508402353

Bates, E. (1991). A functionalist approach to language and its implications for assessment and intervention, In T. Gallaher (Ed), *Pragmatics of language: Clinical practice issues*. Springer.

Bawden, H. N., Stokes, A., Camfield, C. S., Camfield, P. R. & Salisbury, S. (1998). Peer relationship problems in children with Tourette's disorder or diabetes mellitus. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 39*(5), 663–668. https://doi.org/10. 1017/S0021963098002480

Becker, A., Hagenberg, N., Roessner, V., Woerner, W., & Rothenberger, A. (2004). Evaluation of the self-reported SDQ in a clinical setting: Do self-reports tell us more than ratings by adult informants? *European Child and Adolescent Psychiatry, 13*(Suppl. 2), 17–24.

Berg, E., Bjornram, C., Hartelius, L., Laakso, K., & Johnels, B. (2002). High-level language difficulties in Parkinson's disease. *Clinical Linguistics and Phonetics, 17*(1), 63-80.

Bignell, S., & Cain, K. (2007). Pragmatic aspects of communication and language comprehension in groups of children differentiated by teacher ratings of inattention and hyperactivity. *British Journal of Developmental Psychology, 25*(4), 499–512. https://doi.org/10.1348/026151006X171343

Bishop, D. (1989). *Children's Communication Checklist (CCC).* Pearson.

Bishop, D. (2003). *Children's Communication Checklist (CCC)* (2nd ed.). Pearson.

Bitsko, R. H., Claussen, A. H., Lichstein, J., Lichstein, J., Black, L. I., Jones, S. E., Danielson, M. L., Hoenig, J. M., Davis Jack, S. P., Brody, D. J., Gyawali, S., Maenner, M. J., Warner, M., Holland, K. M., Perou, R., Crosby, A. E., Blumberg, S. J., Avenevoli, S., Kaminski, J. W. & Ghandour, R. M. (2022). Mental health among children— United States, 2013–2019. *Morbidity and Mortality Weekly Report, 71*(2), 1–48.

Bloch, M. H. (2013). Clinical course and adult outcome in Tourette syndrome. In: D. Martino & J. F. Leckman (Eds.), *Tourette syndrome* (pp. 107–120). Oxford University Press. https://doi.org/10.1093/med/ 9780199796267.003.0005

Bloch, M. H., Sukhodolsky, D. G., Leckman, J. F., & Schultz, T. (2006). Fine-motor skill deficits in childhood predict adulthood tic severity and global psychosocial functioning in Tourette's syndrome. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 47*(6), 551–559. https://doi.org/10.1111/j.1469-7610.2005.01561.x

Bloom, L., & Lahey, M. (1978). *Language development and language disorders*. New York: John Wiley & Sons.

Borg, A. M., Kaukonen, P., Salmelin, R., Joukamaa, M., & Tamminen, T. (2012). Reliability of the Strengths and Difficulties Questionnaire among Finnish 4-9-year-old children. *Nordic Journal of Psychiatry, 66*(6), 403–413. https://doi.org/10.3109/08039488.2012.660706

Bornstein, R.A. (1990). Neuropsychological performance in children with Tourette's syndrome. *Psychiatry Research, 33*(1), 73–81. https://doi.org/10.1016/0165-1781(90)90150-4

Bornstein, R. A., Baker, G. B., Bazylewich, T. & Douglass, A. B. (1991). Tourette syndrome and neuropsychological performance. *Acta Psychiatrica Scandinavica, 84*(3), 212–216. https://doi.org/10.1111/ j.1600-0447.1991.tb03131.x

Bornstein, M. H., Hahn, C. S., & Suwalsky, J. T. D. (2013). Language and internalizing and externalizing behavioral adjustment: Developmental pathways from childhood to adolescence. *Development and Psychopathology, 25*(3), 857–878. https://doi.org/10.1017/S0954579413000217

Bornstein, R. A., King, G. & Carroll, A. (1983) Neuropsychological abnormalities in Gilles de la Tourette's syndrome. The Journal of Nervous and Mental Disease, 171(8), 497–502.

Bourdon, K. H., Goodman, R., Rae, D. S., Simpson, G., & Koretz, D. S. (2005). The Strengths and Difficulties Questionnaire: U.S. normative data and psychometric properties. *Journal of the American Academy of Child and Adolescent Psychiatry, 44*(6), 557–564. https://doi.org/10.1097/01.chi.0000159157.57075.c8

Brand, N., Geenen, R., Oudenhoven, M., Lindenborn, B., Van Der Ree, A., Cohen-Kettenis, P. & Buitelaar, J. K. (2002). Brief report: Cognitive functioning in children with Tourette's

syndrome with and without comorbid ADHD. *Journal of Pediatric Psychology, 27*(2), 203–208. https://doi.org/10.1093/jpepsy/27.2.203

Braun, V., & Clarke, V. (2021). Can I use TA? Should I use TA? Should I not use TA? Comparing reflexive thematic analysis and other pattern-based qualitative analytic approaches. *Counselling and Psychotherapy Research, 21*(1), 37–47. https://doi.org/10.1002/capr.12360

Braun, V., & Clarke, V. (2022). Thematic analysis: A practical guide. Sage.

Bronfenbrenner, U. (1979). *The ecology of human development: Experiments by nature and design*. Harvard University Press.

Brookshire, B. L., Butler, I. J., Ewing-cobbs, L., & Fletcher, J.M. (1994). Neuropsychological characteristics of children with Tourette syndrome: Evidence for a nonverbal learning disability. *Journal of Clinical and Experimental Neuropsychology, 16*(2), 289–302. https://doi.org/10.1080/01688639408402639

Burd, L., Christensen, T. & Kerbeshian, J. (2008). Speech, language, and communication in Tourette's syndrome. *Annual Review of Applied Linguistics, 28*, 170–190. https://doi.org/10.1017/ S026719050808001X

Burd, L., Freeman, R. D., Klug, M. G. & Kerbeshian, J. (2005). Tourette syndrome and learning disabilities. *BMC Pediatrics, 5*(34), 7–12. https://doi.org/10.1186/1471-2431-5-34

Carroll, L. J., & Rothe, J. P. (2010). Levels of reconstruction as complementarity in mixed methods research: A social theory-based conceptual framework for integrating qualitative and quantitative research. *International Journal of Environmental Research and Public Health, 7*(9), 3478–3488. https://doi.org/10.3390/ijerph7093478

Carrow-Woolfolk, E. (2017). *The Comprehensive Assessment of Spoken Language (CASL-2)* (2nd ed.). WPS.

Carter, A. S., O'Donnell, D. A., Scahill, L., Schultz, R. T., Leckman, J. F., & Pauls, D. L. (2000). Social and emotional adjustment in children affected with Gilles de la Tourette's Syndrome: Associations with ADHD and family functioning. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 41*(2), 215–223. https://doi.org/10.1017/S0021963099005156

Cath, D. C., Muller-Vahl, K. R., & Mathews, C. A. (2022). Other psychiatric comorbidities in Tourette syndrome. In D. Martino & J. F. Leckman (Eds.), *Tourette syndrome* (2nd ed., pp. 65–94). Oxford University Press.

Cavanna, A. E., Luoni, C., Selvini, C., Blangiardo, R., Eddy, C. M., Silvestri, P. R., Cali', P. V., Gagliardi, E., Balottin, U., Cardona, F., Rizzo, R., & Termine, C. (2013). Parent and self-report health-related quality of life measures in young patients with Tourette syndrome. *Journal of Child Neurology, 28*(10), 1305–1308. https://doi.org/10.1177/0883073812457462

Champion, L. M., Fulton, W. A. & Shady, G. A. (1988). Tourette syndrome and social functioning in a Canadian population. *Neuroscience and Biobehavioral Reviews, 12*(3–4), 255–257. https://doi. org/10.1016/S0149-7634(88)80054-X

Chang, S. W., McCracken, J. T., & Piacentini, J. C. (2007). Neurocognitive correlates of child obsessive compulsive disorder and Tourette syndrome. *Journal of Clinical and Experimental Neuropsychology, 29*(7), 724–733. https://doi.org/10.1080/13825580600966383

Channon, S., Crawford, S., Vakili, K. & Robertson, M. M. (2003a). Real-life-type problem solving in Tourette syndrome. *Cognitive and Behavioral Neurology, 16*(1), 3–15. https://doi.org/10.1097/00146965-200303000-00002

Channon, S., Drury, H., Gafson, L., Stern, J., & Robertson, M. M. (2012). Judgements of social inappropriateness in adults with Tourette's syndrome. *Cognitive Neuropsychiatry, 17*(3), 246–261. https://doi.org/10.1080/13546805.2011.590689

Channon, S., Pratt, P. & Robertson, M. M. (2003b). Executive function, memory, and learning in Tourette's syndrome. *Neuropsychology, 17*(2), 247–254. https://doi.org/10.1037/0894-4105.17.2.247

Channon, S., Sinclair, E., Waller, D., Healey, L., & Robertson, M. M. (2004). Social cognition in Tourette's syndrome: Intact theory of mind and impaired inhibitory functioning. *Journal of Autism and Developmental Disorders, 34*(6), 669–677. https://doi.org/10.1007/s10803-004-5287-x

Chowdhury, U., & Christie, D. (2002). Focus on practice: Tourette syndrome: A training day for teachers. *British Journal of Special Education, 29*(3), 123–126. https://doi.org/10.1111/1467-8527.00254

Church, J. A., Wenger, K. K., Dosenbach, N. U. F., Miezin, F. M., Petersen, S. E., & Schlaggar, B. L. (2009). Task control signals in pediatric Tourette syndrome show evidence of immature and anomalous functional activity. *Frontiers in Human Neuroscience, 3*, 1–14. https://doi.org/10.3389/neuro.09.038.2009

Claussen, A. H., Bitsko, R. H., Holbrook, J. R., Bloomfield, J., & Giordano, K. (2018). Impact of Tourette syndrome on school measures in a nationally representative sample. *Journal of*

Developmental and Behavioral Pediatrics, 39(4), 335–342. https://doi.org/10.1097/DBP.0000000000000550.

Climie, E. A., & Mastoras, S. M. (2015). ADHD in schools: Adopting a strengths-based perspective. Canadian Psychology, 56(3), 295–300. https://doi.org/10.1037/cap0000030

Cole, D. (2001). Narrative development in aggressive boys. *Behavioral Disorders, 26*(4), 332–343. https://doi.org/10.1177/019874290102600401

Comings, D. E. & Comings, B. G. (1991). Clinical and genetic relationships between autism-pervasive developmental disorder and Tourette syndrome: A study of 19 cases. *American Journal of Medical Genetics, 39*(2), 180–191. https://doi.org/10.1002/ajmg. 1320390213

Constantino, J. N., & Gruber, C. P. (2012). *Social Responsiveness Scale (SRS)* (2nd ed.). Western Psychological Services.

Cooper, C., Robertson, M. M., & Livingston, G. (2003). Psychological morbidity and caregiver burden in parents of children with Tourette's disorder and psychiatric comorbidity. *Journal of the American Academy of Child & Adolescent Psychiatry, 42*, 1370– 1375. https://doi.org/10.1097/01.CHI.0000085751.71002.48

Cravedi, E., Deniau, E., Giannitelli, M., Pellerin, H., Czernecki, V., Priou, T., Xavier, J., Consoli, A., Hartmann, A., & Cohen, D. (2018). Disentangling Tourette syndrome heterogeneity through hierarchical ascendant clustering. *Developmental Medicine and Child Neurology, 60*(9), 942–950. https://doi.org/10.1111/dmcn.13913.

Creswell, J. W., & Plano Clark, V. (2018). *Designing and conducting mixed methods research* (3rd ed.). Sage.

Creswell, J. W., & Poth, C. N. (2018). *Qualitative inquiry and research design: Choosing among five approaches.* Sage.

Crotty, M. (1998). *The foundations of social research: Meaning and perspectives in the research process.* Sage.

Cullen, B., Samuels, J., Grados, M., Landa, R., Bienvenu, O. J., Liang, K. Y., Riddle, M., Hoehn-Saric, R., & Nestadt, G. (2008). Social and communication difficulties and obsessive-compulsive disorder. *Psychopathology, 41*(3), 194–200. https://doi.org/10.1159/000120988

Cumming, T. M., Strnadová, I., Danker, J., & Basckin, C. (2020). "I was taught that my being was inherently wrong": Is applied behavioural analysis a socially valid practice? *International Journal of Arts Humanities and Social Sciences Studies, 5*(12), 72–82. https://doi.org/10.1007/s40614-022-00364-9

Dall, M., Fellinger, J., & Holzinger, D. (2022). The link between social communication and mental health from childhood to young adulthood: A systematic review. *Frontiers in Psychiatry, 13*(October). https://doi.org/10.3389/fpsyt.2022.944815

Darrow, S. M., Hirschtritt, M. E., Davis, L. K., Illmann, C., Osiecki, L., Grados, M., Sandor, P., Dion, Y., King, R., Pauls, D., Budman, C. L., Cath, D. C., Greenberg, E. Lyon, G. J., Yu, D, McGrath, L. M., McMahon, W. M., Lee, P. C., Delucchi, K. L… & Mathews, C. A. (2017). Autism spectrum symptoms in a Tourette's disorder sample. *Journal of the American Academy of Child and Adolescent Psychiatry, 56*(7), 610–617.

Daub, O., Cunningham, B. J., Bagatto, M. P., Johnson, A. M., Kwok, E. Y., Smyth, R. E., & Cardy, J. O. (2021). Adopting a conceptual validity framework for testing in speech-language pathology. *American Journal of Speech-Language Pathology, 30*(4), 1894–1908. https://doi.org/10.1044/2021_AJSLP-20-00032

De Groot, C. M., Yeates, K. O., Baker, G. B., & Bornstein, R. A. (1997). Impaired neuropsychological functioning in Tourette's syndrome subjects with co-occurring obsessive-compulsive and attention deficit symptoms. *Journal of Neuropsychiatry and Clinical Neurosciences, 9*(2), 267–272. https://doi.org/10.1176/jnp.9.2.267

de Jong, T. J., van der Schroeff, M. P., Achterkamp, M. D., & Vroegop, J. L. (2023). First results of the Strengths and Difficulties Questionnaire, applied as a screening tool for psychosocial difficulties in pediatric audiology. *European Archives of Oto-Rhino-Laryngology, 280*(10), 4467-4476. https//:doi.org/10.1007/s00405-023-07979-x

De Nil, L. F., Roth, I. D., Saltzman-Benaiah, J. & Sandor, P. (2006). Speech, language, and learning deficiencies associated with Tourette's syndrome. *Perspectives on Neurophysiology and Neurogenic Speech and Language Disorders, 16*(2), 12–16. https://doi.org/10.1044/nnsld16.2.12

De Nil, L. F., Sasisekaran, J, Van Lieshout, P. H. H. M., & Sandor, P. (2005). Speech disfluencies in individuals with Tourette syndrome. *Journal of Psychosomatic Research, 58*(1), 97–102.

Devinsky, O., Bear, D., Moya, K., & Benowitz, L. (1993). Perception of emotion in patients with Tourette's syndrome. *Neuropsychiatry, Neuropsychology and Behavioral Neurology, 6*(3), 166–169.

Donaher, J. (2006). Tourette's syndrome and stuttering. *Perspectives on Fluency and Fluency Disorders, 16*(2), 5–6. https://doi.org/10.1044/ ffd16.2.5

Donaher, J. (2008). *Speech fluency demonstrated by children with Tourette syndrome* (3320012) [Doctoral Dissertation, Temple University]. https://doi.org/10.1016/b978-012397720-5.50034-7

Drury, H., Channon, S., Barrett, R., Young, M. B., Stern, J. S., Simmons, H., & Crawford, S. (2012). Emotional processing and executive functioning in children and adults with Tourette's syndrome. *Child Neuropsychology, 18*(3), 281–298. https://doi.org/10.1080/09297049.2011.613811.

Drury, H., Shah, S., Stern, J. S., Crawford, S., & Channon, S. (2018). Comprehension of direct and indirect sarcastic remarks in children and adolescents with Tourette's syndrome. *Child Neuropsychology, 24*(4), 490–509.

Dye, C. D., Walenski, M., Mostofsky, S. H., & Ullman, M. T. (2016). A verbal strength in children with Tourette syndrome? Evidence from a non-word repetition task. *Brain and Language, 160*, 61–70. https://doi.org/10.1016/j.bandl.2016.07.005

Dykens, E., Leckman, J., Riddle, M., Hardin, M., Schwartz, S. & Cohen, D. (1990). Intellectual, academic, and adaptive functioning of Tourette syndrome children with and without attention deficit disorder. *Journal of Abnormal Child Psychology, 18*(6), 607–615. https://doi.org/10.1007/BF01342750

Eapen, V., Cavanna, A. E., & Robertson, M. M. (2016). Comorbidities, social impact, and quality of life in Tourette syndrome. Frontiers in psychiatry, 7, 1-6.

Eapen, V., McPherson, S., Karlov, L., Nicholls, L., Črnčec, R. & Mulligan, A. (2019). Social communication deficits and restricted repetitive behavior symptoms in Tourette syndrome. *Neuropsychiatric Disease and Treatment, 15*, 2151–2160. https://doi.org/10.2147/ ndt.s210227

Eddy, C. M. (2021). Tourette syndrome. In L. Cummings (Ed.), *Handbook of Pragmatic Language Disorders: Complex and Underserved Populations* (pp. 375-383). Springer.

Eddy, C. M., & Cavanna, A. E. (2013). Altered social cognition in Tourette syndrome: Nature and implications. *Behavioural Neurology, 27*(1), 15–22. https://doi.org/10.3233/BEN-120298

Eddy, C.M. & Cavanna, A.E. (2015) Triangles, tricks and tics: hypermentalizing in response to animated shapes in Tourette syndrome. *Cortex, 71*, 68–75.

Eddy, C. M., Cavanna, A. E., & Hansen, P. C. (2017). Empathy and aversion: the neural signature of mentalizing in Tourette syndrome. *Psychological Medicine, 47*(3), 507–517.

Eddy, C.M., Cavanna, A.E., Rickards, H.E. & Hansen, P.C. (2016) Temporo-parietal dysfunction in Tourette syndrome: insights from an fMRI study of theory of mind. *Journal of Psychiatric Research, 81*, 102–11.

Eddy, C. M., Macerollo, A., Martino, D., & Cavanna, A. E. (2015). Interpersonal reactivity differences in Tourette syndrome. *Psychiatry Research, 228*(3), 932–935. https://doi.org/10.1016/j.psychres.2015. 05.070

Eddy, C. M., Mitchell, I. J., Beck, S. R., Cavanna, A. E., & Rickards, H. E. (2010a). Altered attribution of intention in Tourette's syndrome. *Journal of Neuropsychiatry and Clinical Neurosciences, 22*(3), 348–351. https://doi.org/10.1176/jnp.2010.22.3.348

Eddy, C. M., Mitchell, I. J., Beck, S. R., Cavanna, A. E., & Rickards, H. E. (2010b). Impaired comprehension of nonliteral language in Tourette syndrome. *Cognitive and Behavioral Neurology, 23*(3), 178–184.

Eddy, C. M., Mitchell, I. J., Beck, S. R., Cavanna, A. E., & Rickards, H. (2011). Social reasoning in Tourette syndrome. *Cognitive Neuropsychiatry, 16*(4), 326–47.

Ehlers, S., Gillberg, C., Wing, L. (1999). A screening questionnaire for Asperger syndrome and other high-functioning autism spectrum disorders in school age children. *Journal of Autism and Developmental Disorders, 29*, 12–141.

Feehan. A, & Charest, M. (2021). A scoping review of oral language and social communication abilities in children with Tourette syndrome. *International Journal of Language and Communication Disorders, 59*(1), 1–22. https://doi.org/10.1111/1460-6984.12949

Ferrari, M., Matthews, W. S. & Barabas, G. (1984). Children with Tourette syndrome: Results of psychological tests given prior to drug treatment. *Developmental and Behavioural Pediatrics, 5*(3), 116–119. https://doi.org/10.1097/00004703-198406000-00002

Fetters M. D. (2020). *The mixed methods research workbook: Activities for designing, implementing, and publishing studies*. Sage.

Fetters, M. D. (2022). A comprehensive taxonomy of research designs, a scaffolded design figure for depicting essential dimensions, and recommendations for achieving design naming conventions in the field of mixed methods research. *Journal of Mixed Methods Research, 16*(4), 394-411. https://doi.org/10.1177/15586898221131238

Fisher, A. P., Gies, L. M., Chapman, L., Aguilar, J. M., Yeates, K. O., Taylor, H. G., & Wade, S. L. (2022). The clinical utility of the Children's Communication Checklist-2 in children with early childhood traumatic brain injury. *Clinical Neuropsychologist, 36*(7), 1728–1745. https://doi.org/10.1080/13854046.2020.1866675

Fitcher, B. (2014). *Homeschooled with Tourette's*. https://mollygreen.com/blog/homeschooled-with-tourettes/

Fletcher, J., Dimitropoulos, G., Martino, D., Wilcox, G., MacMaster, F., Arnold, P., & Pringsheim, T. (2021). Developing a provincial patient support network for children and

families affected by Tourette syndrome and/or obsessive–compulsive disorder: Results of a stakeholder consultation. *Child and Adolescent Psychiatry and Mental Health, 15*(1), 1–10. https://doi.org/10.1186/s13034-021-00383-5

Gaddy, C., & Crow, H. (2023). A primer on neurodiversity-affirming speech and language services for autistic individuals. *Perspectives of the ASHA Special Interest Groups, 8*(6), 1220-1237. https://doi.org/10.1044/2023_persp-23-00106

Gadow, K.D. & Paolicelli, L. M. (1986). *The Global Tic Rating Scale (GTRS).* State University of New York Department of Psychiatry.

Gallina, N. B. (1989). *Tourette's Syndrome children: Significant achievement and social behavior variables* (Publication No. 9000024) [Doctoral dissertation, City University of New York]. ProQuest Dissertations and Theses Global.

Ghahari, S., Hassani, H., & Purmofrad, M. (2017). Pragmatic competency and obsessive–compulsive disorder: A comparative assessment with normal controls. *Journal of Psycholinguistic Research, 46*(4), 863–875. https://doi.org/10.1007/s10936-016-9467-6

Ghanizadeh, A., Mohammadi, M. R., & Dehbozorgi, G. R. (2010). Children and their parent's perceptions of symptom severity and treatment preference for Tourette syndrome. *Iranian Journal of Psychiatry, 5*(3), 93–936.

Gilkey-Hirn, R., & Park, K. L. (2012). Teacher-mediated instructional strategies for students with emotional or behavioural disorders. *Beyond Behaviour*, (Fall), 1–9.

Gillam, R. B., & Pearson, N. A. (2017). *Test of Narrative Language (TNL)* (2nd ed.). WPS.

Goodman, R. (1997). *Strengths and Difficulties Questionnaire (SDQ)* [Database record].

Goodman, R., Renfrew, D., & Mullick, M. (2000). Predicting type of psychiatric disorder from Strengths and Difficulties Questionnaire (SDQ) scores in child mental health clinics in London and Dhaka. *European Child and Adolescent Psychiatry, 9*, 129–134.

Goodman, R., Ford, T., Corbin, T., & Meltzer, H. (2004). Using the Strengths and Difficulties Questionnaire (SDQ) multi-informant algorithm to screen looked-after children for psychiatric disorders. *European Child and Adolescent Psychiatry, 13*, 25–31.

Goodman R., & Scott, S. (1999). Comparing the Strengths and Difficulties Questionnaire and the Child Behavior Checklist: Is small beautiful? *Journal of Abnormal Child Psychology, 27*, 17–24.

Gorman, D. A., Thompson, N., Plessen, K. J., Robertson, M. M., Leckman, J.F., & Peterson, B. S. (2010). Psychosocial outcome and psychiatric comorbidity in older adolescents with Tourette syndrome: controlled study. *British Journal of Psychiatry, 197*(1), 36–44. https://doi.org/10.1192/bjp.bp.109.071050

Greene, D. J., Black, K. J., & Schlaggar, B. L. (2013). Neurobiology and Functional Anatomy of Tic Disorders. In: D. Martino & J. F. Leckman (Eds., pp. 238–275). *Tourette Syndrome*. Oxford University Press. https://doi. org/10.1093/med/9780199796267.003.0012

Güler, A. S., Berkem, M., Yazgan, Y., & Kalaça, S. (2015). Cognitive flexibility and social responsiveness in children and adolescents with Tourette syndrome. *Child Psychiatry and Human Development, 46*(6), 940–950. https://doi.org/10.1007/s10578-015-0533-3.

Gutierrez-Colina, A. M., LaMotte, J., Eaton, C., Kardon, P., & Blount, R. L. (2015). The role of self-competence in health-related quality of life and behavioral functioning of children with Tourette syndrome. *Journal of Developmental and Behavioral Pediatrics, 36*(9), 743–751. https://doi.org/10.1097/DBP.0000000000000214

Hagin, R. A., & Kugler, J. (1998). School problems associated with Tourette syndrome. In: D. J. Cohen, R. D. Bruun, & J. F. Leckman (Eds.), *Tourette's syndrome and tic disorders: Clinical understanding and treatment* (pp. 223–236). John Wiley.

Halls, G., Cooper, P. J., & Creswell, C. (2015). Social communication deficits: Specific associations with social anxiety disorder. *Journal of Affective Disorders, 172*, 38–42. https://doi.org/10.1016/j.jad.2014.09.040

Harris, E. L., Schuerholz, L. J., Singer, H. S., Reader, M. J., Brown, J., Cox, C., Mohr, J., Chase, G. A. & Denckla, M. B. (1995). Executive function in children with Tourette Syndrome and/or Attention Deficit Hyperactivity Disorder. *Journal of the International Neuropsychological Society, 1*, 511–516. https://doi.org/10.1016/j.cortex. 2019.11.007

Harrison, H., Birks, M., Franklin, R., & Mills, J. (2017). Case study research: Foundations and methodological orientations. *Forum Qualitative Sozialforschung, 18*(1). https://doi.org/10.17169/fqs-18.1.2655

Helland, W. A., Helland, T., & Heimann, M. (2014) Language profiles and mental health problems in children with specific language impairment and children with ADHD. *Journal of Attention Disorders, 18*(3), 226-235. https://doi.org/10.1177/1087054712441705

Hoekstra, P. J., & Dietrich, A., (2022). Perinatal adversities and Tourette syndrome. In D. Martino & J. F. Leckman (Eds.), *Tourette syndrome* (2nd ed., pp. 1-25). Oxford University Press.

Hoekstra, P. J., Steenhuis, M. P., Troost, P. W., Korf, J., Kallenberg, C. G. M. & Minderaa, R. B. (2004). Relative contribution of attention deficit hyperactivity disorder, obsessive-compulsive disorder, and tic severity to social and behavioral problems in tic disorders.

Journal of Developmental and Behavioral Pediatrics, 25(4), 272–279. https://doi.org/10.1097/00004703-200408000-00007

Hollo, A., Wehby, J. H., & Oliver, R. M. (2013). Unidentified language deficits in children with emotional and behavioral disorders: A meta-analysis. *Exceptional Children, 80*(2), 169–186. https://doi.org/10.1177/001440291408000203

Houghton, C., Murphy, K., Shaw, D., Casey, D. (2015). Qualitative case study data analysis: An example from practice. *Nurse Researcher, 22*(5), 8-12. doi: 10.7748/nr.22.5.8.e1307. PMID: 25976531.

Huang, Y. (2015). *Pragmatics* (2nd ed.). Oxford University Press.

Hulbert, T. (1986). *Neuropsychological subgrouping of Tourette children* [Unpublished doctoral dissertation]. Wayne State University. https://doi.org/10.1017/CBO9781107415324.004

Hyter, Y. (2017). Pragmatic assessment and intervention in children. In L. Cummins (Ed.), *Research in Clinical Pragmatics* (pp. 493-526). Springer International Publishing.

Hyter, Y. (2007). Pragmatic language assessment: A pragmatics-as-social practice model. *Topics in Language Disorders, 27*(2), 128-145.

Incagnoli, T. & Kane, R. (1983). Developmental perspective of the Gilles de la Tourette syndrome. *Perceptual and Motor Skills, 57*, 1271–1281. https://doi.org/10.2466/pms.1983.57.3f.1271

Jeans, A. D. (2016). *The school and social experiences of Tourette syndrome through the first-person perspective of youth: A theatre-based approach to community-based research* [Master's Thesis]. University of Alberta.

Jensen, J. (2004). Neuropsychological and behavioural characteristics of children and adolescents with Tourette Syndrome compared to controls with dyslexia. *Journal of Allergy and Clinical Immunology, 130*(2), 556. https://doi.org/10.1016/j.jaci.2012.05.050

Kadesjö, B. & Gillberg, C. (2000). Tourette's disorder: Epidemiology and comorbidity in primary school children. *Journal of the American Academy of Child and Adolescent Psychiatry, 39*(5), 548–555. https://doi.org/10.1097/00004583-200005000-00007

Kalfayan Sweeten, K. J. (1997). *Developmental issues in male adolescents with Tourette's syndrome.* [Unpublished doctoral dissertation]. California School of Professional Psychology.

Katsovich, L., Findley, D., & Leckman, J.F. (2003). Disruptive behavior in children with Tourette's syndrome: Association with ADHD comorbidity, tic severity, and functional impairment. *Journal of the American Academy of Child and Adolescent Psychiatry, 42*(1), 98–105. https://doi.org/10.1097/00004583- 200301000-00016

Kelso, K. (2012). *Comparing the Children's Communication Checklist to Standardized Tests: Results from School-age Children Adopted from Haiti* [Unpublished Report].

Khalifa, N., Dalan, M. & Rydell, A. M. (2010). Tourette syndrome in the general child population: Cognitive functioning and self-perception. *Nordic Journal of Psychiatry, 64*(1), 11–18. https://doi.org/ 10.3109/08039480903248096

Khalil, H., Peters, M., Godfrey, C.M., McInerney, P., Soares, C. B. & Parker, D. (2016). An evidence-based approach to scoping reviews. *Worldviews on Evidence-Based Nursing, 13*(2), 118–123. https://doi. org/10.1111/wvn.12144

Kjærandsen, K. S., Handegård, B. H., Brøndbo, P. H., & Halvorsen, M. B. (2023). Impact supplement of the Strengths and Difficulties Questionnaire in the assessment of

functional impairment in children with ADHD or ASD in a mixed neuropediatric sample: A partial validation study. *Journal of Mental Health Research in Intellectual Disabilities, 16*(1), 1-22. https//:doi.org/10.1080/19315864.2022.2148788

Klasen, H., Woerner, W., Wolke, D., Meyer, R., Overmeyer, S., Kaschnitz, W., Rothenberger, A., & Goodman, R. (2000). Comparing the German versions of the Strengths and Difficulties Questionnaire (SDQ-Deu) and the Child Behavior Checklist. *European Child and Adolescent Psychiatry, 9*, 271–276.

Korrel, H., Mueller, K. L., Silk, T., Anderson, V., & Sciberras, E. (2017). Research Review: Language problems in children with Attention-Deficit Hyperactivity Disorder: A systematic meta-analytic review. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 58*(6), 640–654. https://doi.org/10.1111/jcpp.12688

Krah, B. (2002). Tourette syndrome: The anatomy of a tic: From the point of view of a person with TS. *ASHA Leader, 7*(14), 5–6. https:// doi.org/10.1044/leader.FTR2.07142002.5

Kwak, C. & Jankovic, J. (2002). Tourette syndrome: The neurology of a tic: From the point of view of the scientist. *ASHA Leader, 7*(14), 5–7. https://doi.org/10.1044/leader.FTR1.07142002.4

Langdon, R., & Coltheart, M. (2004). Recognition of metaphor and irony in young adults: The impact of schizotypal personality traits. *Psychiatry Research, 125*(1), 9-20.

Lanser, J. B. K., Van Santen, W. H. C., Jennekens-Schinkel, A. & Roos, R. A. C. (1993). Tourette's syndrome and right hemisphere dysfunction. *British Journal of Psychiatry, 163*, 116–118. https://doi.org/10. 1192/bjp.163.1.116

Leckman, J. F., Bloch, M. H., Sukhodolsky, D. G., Artukoglu, B. B., Scahill, L., & King, R. A. (2022). Phenomenology of tics and sensory urges. In D. Martino & J. F. Leckman (Eds.), *Tourette syndrome* (2nd ed., pp. 1-25). Oxford University Press.

Lee, M. Y., Mu, P. F., Wang, W. S., & Wang, H. S. (2016). "Living with tics": Self-experience of adolescents with Tourette syndrome during peer interaction. *Journal of Clinical Nursing, 25*(3–4), 463–471. https://doi.org/10.1111/jocn.13074.

Legg, C., Penn, C., Temlett, J., & Sonnenberg, B. (2005). Language skills of adolescents with Tourette Syndrome. *Clinical Linguistics and Phonetics, 19*(1), 15–33. https://doi.org/10.1080/02699200410001665270.

Lethlian, J. B., & Murdoch, B. E. (1997). Performance of subjects with multiple sclerosis on tests of high level language. *Aphasiology, 11*(1), 39-57.

Levac, D., Colquhoun, H. & O'Brien, K. K. (2010). Scoping studies: Advancing the methodology. *Implementation Science, 5*(1), 1–10. https://doi.org/10.1186/1748-5908-5-69

Lincoln, Y. S., & Guba E. G. (1985). *Naturalistic Inquiry*. SAGE.

Ludlow, C. L., Polinsky, R. J., Caine, E. D., Bassich, C. J. & Ebert, M. H. (1982). Language and speech abnormalities in Tourette syndrome. In A. J. Friedhoff & T. N. Chase (Eds.), *Gilles de la Tourette syndrome* (pp. 351–361). Raven Press.

MacKie, L., & Law, J. (2010). Pragmatic language and the child with emotional/behavioural difficulties (EBD): A pilot study exploring the interaction between behaviour and communication disability. *International Journal of Language and Communication Disorders, 45*(4), 397–410. https://doi.org/10.3109/13682820903105137

Mahone, E. M., Cirino, P. T., Cutting, L. E., Cerrone, P. M., Hagelthorn, K. M., Hiemenz, J. R., Singer, H. S. & Denckla, M. B. (2002). Validity of the behavior rating inventory of executive function in children with ADHD and/or Tourette syndrome. *Archives of Clinical Neuropsychology, 17*(7), 643–662. https://doi.org/10.1016/S0887-6177(01)00168-8

Mahone, E. M., Koth, C. W., Cutting, L., Singer, H. S. & Denckla, M. B. (2001). Executive function in fluency and recall measures among children with Tourette syndrome or ADHD. *Journal of the International Neuropsychological Society, 7*(1), 102–111. https://doi.org/ 10.1017/S1355617701711101

Matthews, D. (2014). *Pragmatic development in first language acquisition.* John Benjamins Publishing Company.

Malli, M. A., Forrester-Jones, R., & Murphy, G. (2016). Stigma in youth with Tourette's syndrome: A systematic review and synthesis. *European Child and Adolescent Psychiatry, 25*(2), 127-139. https://doi.org/10.1007/s00787-015-0761-x

Marek, E. (2006). *Social functioning and quality of life for individuals with Tourette's Syndrome.* [Unpublished Doctoral Dissertation]. The Chicago School of Professional Psychology. https://doi.org/10. 1016/j.jaci.2012.05.050

Marks, K. P., Pouwer, F., Jensen, M. B., Ibfelt, E. H., Kristensen, L. J., & Thastum, M. (2023). Responses to the Strengths and Difficulties Questionnaire predict HbA1c trajectories in children and adolescents with type 1 diabetes: A population-based study. *BMJ Open Diabetes Research and Care, 11*(6) 1-9. Https//:doi.org/10.1136/bmjdrc-2023-003479

Martino, D., & Leckman, J. F. (2013). *Tourette syndrome*. Oxford University Press.

Martino, D., & Leckman, J. F. (2022). *Tourette syndrome* (2nd ed.). Oxford University Press.

Martino, D., Pringsheim, T. M., Cavanna, A. E., Colosimo, C., Hartmann, A., Leckman, J. F., Luo, S., Munchau, A., Goetz, C. G., Stebbins, G. T., Martinez-Martin, P. (2017). Systematic review of severity scales and screening instruments for tics: Critique and recommendations. *Movement Disorders, 32*(3), 467–473. https://doi.org/10.1002/mds.26891

Miles, M. B. & Huberman, A. M. (1994). *Qualitative data analysis: An expanded sourcebook* (2nd ed.). Sage.

Mingbunjerdsuk, D., & Zinner, S. H. (2020). Coping with Tics in School and with Peers. *Current Developmental Disorders Reports, 7*(4), 227–236. https://doi.org/10.1007/s40474-020-00218-6

Morand-Beaulieu, S., Grot, S., Lavoie, J., Leclerc, J. B., Luck, D. & Lavoie, M. E. (2017a). The puzzling question of inhibitory control in Tourette syndrome: A meta-analysis. *Neuroscience and Biobehavioral Reviews, 80*, 240–262. https://doi.org/10.1016/j.neubiorev. 2017.05.006

Morand-Beaulieu, S., Leclerc, J. B., Valois, P., Lavoie, M. E., O'Connor, K. P., & Gauthier, B. (2017). A review of the neuropsychological dimensions of Tourette syndrome. *Brain Sciences, 7*(106). https://doi.org/10.3390/brainsci7080106.

Morse, J. M. (1994). 'Emerging from the data': the cognitive processes of analysis in qualitative inquiry. In J. M. Morse (Ed.), *Critical issues in qualitative research methods.* Sage.

Morse, J. M., Barrett, M., Mayan, M., Olson, K., & Spiers, J. (2002). Verification Strategies for Establishing Reliability and Validity in Qualitative Research. *International Journal of Qualitative Methods, 1*(2), 13–22. https://doi.org/10.1177/160940690200100202

Murphy, T. & Eddy, C. M. (2013). Neuropsychological assessment in Tourette syndrome. In: D. Martino & J. F. Leckman (Eds.), *Tourette syndrome* (pp. 439–467). Oxford University Press.

Norbury, C., Gooch, D., Wray, C., Baird, G., Charman, T., Simonoff, E., Vamvakas, G., & Pickles, A. (2016). The impact of nonverbal ability on prevalence and clinical presentation of language disorder: Evidence from a population study. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 57*(11), 1247-1257.

Nussey, C., Pistrang, N., & Murphy, T. (2014). Does it help to talk about tics? An evaluation of a classroom presentation about Tourette syndrome. *Child and Adolescent Mental Health, 19*(1), 31–38. https://doi.org/10.1111/camh.12000

O'Hare, D., Eapen, V., Helmes, E., McBain, K., Reece, J., & Grove, R. (2015). Factors impacting the quality of peer relationships of youth with Tourette's syndrome. *BMC Psychology, 3*(1), 1–13. https://doi.org/10.1186/S40359-015-0090-3

O'Neill, D. K. (2012). Components of pragmatic ability and children's pragmatic language development. In H. Schmid (Ed.), *Cognitive pragmatics* (pp. 261-287). Hubert & Co.

O'Quinn, A. N. & Thompson, R. J. (1980). Tourette's syndrome: An expanded view. *Pediatrics, 66*(3), 420–424. https://doi.org/10.1016/ s0002-7138(09)61008-6

Oates, M., & Bean, A. (2023). Qualitative Analysis of the Experiences and Perspectives of Autistic Speech-Language Pathologists. *American Journal of Speech-Language Pathology, 32*(5), 2178-2191. https://doi.org/10.1044/2023_AJSLP-22-00399

Parke R. D., Roisman, G. I., & Rose, A. J. (2019). *Social development*. Wiley & Sons.

Paul, R., Norbury, C., & Gosse, C. (2017). *Language disorders from infancy through adolescence: Listening, speaking, reading, writing, and communicating* (5th ed). Elsevier.

Perkins, M. R. (2014). Pragmatics as interaction. In M. J. Ball, N. Müller, & R. L. Nelson (Eds.), *Handbook of qualitative research in communication disorders* (pp. 131–147). Routledge.

Peters, M. D. J., Godfrey, C. M., Khalil, H. McInerney, P., Parker, D. & Soares, C. B. (2015). Guidance for conducting systematic scoping reviews. *International Journal of Evidence-Based Healthcare, 13*(3), 141–146.

Philofsky, A., Fidler, D. J., & Hepburn, S. (2007). Pragmatic language profiles of school-age children with autism spectrum disorders and Williams Syndrome. *American Journal of Speech-Language Pathology, 16*(4), 368–380. https://doi.org/10.1044/1058-0360(2007/040)

Pine, R., Bruckner, K., Mbinta, J., Twemlow, R., & Anderson, S. (2022). Perceptions of parents and caregivers in New Zealand: Educational experiences of their children with Tourette syndrome. *International Journal of Disability, Development and Education, 71*(3), 1–14. https://doi.org/10.1080/1034912X.2022.2120603

Pratt, P. L. (2000). *Executive functioning and habit learning in children with Tourette Syndrome*. [Unpublished Doctoral Dissertation]. University College London.

Pringsheim, T., & Hammer, T. (2013). Social behavior and comorbidity in children with tics. *Pediatric Neurology, 49*(6), 406–410. https://doi.org/10.1016/j.pediatrneurol.2013.08.005

Prutting, C. A., & Kirchner, D. M. (1987). A clinical appraisal of the pragmatic aspects of language. *Journal of Speech and Hearing Disorders, 52*(2), 105–119. https://doi.org/10.1044/jshd.5202.105

Prutting, C. A. (1982). Pragmatics as social competence. *Journal of Speech and Hearing Disorders, 47*, 123–134.

Rajendran, G., Mitchell, P. & Rickards, H. (2005). How do individuals with Asperger syndrome respond to nonliteral language and inappropriate requests in computer-mediated communication? *Journal of Autism and Developmental Disorders, 35*(4), 429–443. https://doi.org/10.1007/s10803-005-5033-z

Redmond, S. M. (2004). Conversational profiles of children with ADHD, SLI and typical development. *Clinical Linguistics and Phonetics, 18*(2), 107–125. https://doi.org/10.1080/02699200310001611612

Richardson, L. (2000). Writing: A method of inquiry. In N. K. Denzin & Y. S. Lincoln (Eds.), *Handbook of qualitative research* (2nd ed., pp. 923-948). Sage.

Ricketts, E. J., Wolicki, S. B., Danielson, M. L., Rozenman, M., McGuire, J. F., Piacentini, J., Mink, J. W., Walkup, J. T., Woods, D. W., Bitsko, R. H. (2022). Academic, interpersonal, recreational, and family impairment in children with Tourette syndrome and Attention-Deficit/Hyperactivity Disorder. *Child Psychiatry and Human Development, 53*(1), 3–15. https://doi.org/10.1007/s10578-020-01111-4

Robertson, M. M., & Eapen, V. (2017). The psychosocial aspects of the Gilles de la Tourette syndrome: Empirical evidence from the literature. *Current Behavioral Neuroscience Reports, 4*(1), 59–69. https://doi.org/10.1007/s40473-017-0107-8

Robinson, L., Bitsko, R., Schieve, L., & Visser, R. (2013). Tourette syndrome, parenting aggravation, and the contribution of co-occurring conditions among a nationally representative sample. *Disability and Health Journal, 6*(1), 26–35. https://doi.org/10.1016/j.dhjo.2012.10.002

Roitsch, J., Phalen, L., & Watson, S. M. R. (2019). Predictors of comfort level in speech-language pathologists working with students with attention-deficit/hyperactivity disorder. *Perspectives of the ASHA Special Interest Groups, 4*(5), 1097-1109. https://doi.org/10.1044/2019_PERS-SIG16-2019-0012

Rothenberger, A., & Roessner, V. (2013). The phenomenology of Attention-Deficit/Hyperactivity Disorder in Tourette syndrome. In D. Martino & J. F. Leckman (Eds.), *Tourette Syndrome* (pp. 439-467). Oxford University Press.

Rutter, M., Bailey, A., & Lord, C. (2003). *The Social Communication Questionnaire (SCQ).* Western Psychological Services.

Samuels, J., Shugart, Y. Y., Wang, Y., Grados, M. A., Bienvenu, O. J., Pinto, A., Rauch, S. L., Greenberg, B. D., Knowles, J. A., Fyer, A. J., Piacentini, J. Pauls, D. L., Cullen, B., Rasmussen, S. A., Stewart, S. E., Geller, D. A., Maher, B. S., Goes, F. S., Murphy, D. L… & Nestadt, G. (2014). Clinical correlates and genetic linkage of social and communication difficulties in families with obsessive-compulsive disorder: Results from the OCD collaborative genetics study. *American Journal of Medical Genetics, Part B: Neuropsychiatric Genetics, 165*(4), 326–336. https://doi.org/10.1002/ajmg.b.32235

Saul, J., Griffiths, S., & Norbury, C. F. (2022). Prevalence and functional impact of social (pragmatic) communication disorders. *Journal of Child Psychology and Psychiatry and Allied Disciplines*, 1–12. https://doi.org/10.1111/jcpp.13705

Scharf, J. M., Miller, L. L., Gauvin, C. A., Alabiso, J., Mathews, C. A., & Ben-Shlomo, Y. (2015). Population prevalence of Tourette syndrome: A systematic review and meta-analysis. *Movement Disorders, 30*(2), 221–228. https://doi.org/10.1002/mds.26089

Scharf, J. M., Miller, L. L., Mathews, C. A., & Ben-Shlomo, Y. (2012). Prevalence of Tourette syndrome and chronic tics in the population-based Avon longitudinal study of parents and children cohort. *Journal of the American Academy of Child and Adolescent Psychiatry, 51*(2), 192-201. https://doi.org/10.1016/j.jaac.2011.11.004

Schoonenboom, J., & Johnson, R. B. (2017). How to construct a mixed methods research design. *Kolner Zeitschrift fur Soziologie und Sozialpsychologie, 69*(Suppl 2), 107–131. https://doi.org/10.1007/s11577-017-0454-1

Schuerholz, L. J., Baumgardner, T. L., Singer, H. S., Reiss, A. L. & Denckla, M. B. (1996). Neuropsychological status of children with Tourette's syndrome with and without attention deficit hyperactivity disorder. *Neurology, 46*(4), 958–965. https://doi.org/10.1212/ WNL.46.4.958

Schuerholz, L. J., Singer, H. S. & Denckla, M. B. (1998). Gender study of neuropsychological and neuromotor function in children with Tourette syndrome with and without attention-deficit hyperactivity disorder. *Journal of Child Neurology, 13*(6), 277–282. https://doi.org/10.1177/088307389801300607

Semel, E., Wiig, E. H., & Secord, W. A. (2003). *Clinical Evaluation of Language Fundamentals (CELF-4)* (4th ed.). Pearson.

Shapiro, E., Shapiro, A. K. & Clarkin, J. (1974). Clinical psychological testing in Tourette's syndrome. *Journal of Personality Assessment, 38*(5), 464–478. https://doi.org/10.1080/00223891.1974.10120005

Shapiro, A. K., Shapiro, E., Young, J. G. & Feinberg, T. E. (1988). *Gilles de la Tourette syndrome* (2nd ed.). Raven Press.

Sherman, E. M. S., Shepard, L., Joschko, M., & Freeman, R. D. (1998). Sustained attention and impulsivity in children with Tourette syndrome: Comorbidity and confounds. *Journal of Clinical and Experimental Neuropsychology, 20*(5), 644–657. https://doi.org/10.1076/jcen.20.5.644.1118

Sikora, K., Roelofs, A., Hermans, D., & Knoors, H. (2019). Executive control in language production by children with and without language impairment. *International Journal of Language and Communication Disorders, 54*(4), 645–655. https://doi.org/10.1111/1460-6984.12470

Singer, H. S. (2013). The Neurochemistry of Tourette Syndrome. In: D. Martino & J. F. Leckman (Eds.), *Tourette syndrome* (pp. 276–300). Oxford University Press. https://doi.org/10.1093/med/9780199796267.003.0013

Smith, H., Fox, J. R. E., & Trayner, P. (2015). The lived experiences of individuals with Tourette syndrome or tic disorders: A meta-synthesis of qualitative studies. *British Journal of Psychology, 106*(4), 609–634. https://doi.org/10.1111/bjop.12118.

Sparrow, S. S., Balla, D. A., & Cicchetti, D. V. (1984). *The Vineland adaptive behavior scales*. American Guidance Service.

Spencer, T., Biederman, J., Harding, M., O'Donnell, D., Wilens, T., Faraone, S., Coffey, B., & Geller, D. (1998). Disentangling the overlap between Tourette's disorder and ADHD. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 39*(7), 1037–1044. https://doi.org/10.1017/S0021963098002984

Stake, R. E. (2006). *Multiple Case Study Analysis*. The Guilford Press.

Stake, R. E. (1995). *The Art of Case Study Analysis*. SAGE.

Statistics Canada (2023b). *Changing demographics of racialized people in Canada*. https://www150.statcan.gc.ca/n1/pub/36-28-0001/2023008/article/00001-eng.htm

Statistics Canada (2023a). *English–French bilingualism in Canada: Recent trends after five decades of official bilingualism*. https://www12.statcan.gc.ca/census-recensement/2021/as-sa/98-200-X/2021013/98-200-X2021013-eng.cfm

Statistics Canada (2016). *More post-secondary education indicators*. https://www150.statcan.gc.ca/n1/en/subjects/education_training_and_learning/postsecondary_education

Stewart, S., Greene, D., Lessov-Schlaggar, C., Church, J., & Schlaggar, B. (2015). Clinical correlates of parenting stress in children with Tourette syndrome and in typically developing children. *The Journal of Pediatrics, 166*(5), 1297–1302e3. https://doi.org/10.1016/j.jpeds.2015.01.041

Stokes, A., Bawden, H. N., Camfield, P. R., Backman, J. E. & Dooley, M. (1991). Peer problems in Tourette's disorder. *Pediatrics, 87*(6), 936– 942.

Suh, H. W., Yoon, S. I., Hong, S., Lee, H. W., Lee, M., Kim, J. W., & Chung, S. Y. (2022). How do children with Tourette's syndrome and their caregivers live with the disorder? A systematic review of qualitative evidence. *Frontiers in Psychiatry, 13*(September), 1–16. https://doi.org/10.3389/fpsyt.2022.992905

Sukhodolsky, D. G., Scahill, L., Zhang, H., Peterson, B. S., King, R. A., Lombroso, P. J., Katsovich, L., Findley, D., & Leckman, J. F. (2003). Disruptive behavior in children with Tourette's syndrome: Association with ADHD comorbidity, tic severity, and functional impairment. *Journal of the American Academy of Child and Adolescent Psychiatry, 42*(1), 98–105. https://doi.org/10.1097/00004583-200301000-00016

Sukhodolsky, D. G., Scahill, L., Zhang, H., Peterson, B. S., King, R. A., Lombroso, P. J., & Thompson, R. J., O'Quinn, A. N., & Logue, P. E. (1979). Gilles de la Tourette syndrome: A review and neuropsychological aspects of four cases. *Journal of Pediatric Psychology, 74*(4), 371–387. https:// doi.org/10.1177/014107688607900508

Takács, Á., Kóbor, A., Chezan, J., Éltető, N., Tárnok, Z., Nemeth, D., Ullman, M. T., & Janacsek, K. (2018). Is procedural memory enhanced in Tourette syndrome? Evidence from a sequence learning task. *Cortex, 100*, 84–94. https://doi.org/10.1016/j.cortex.2017.08.037

Timler, G. R. (2014). Use of the Children's Communication Checklist—2 for classification of language impairment risk in young school-age children with Attention-Deficit/Hyperactivity Disorder. *American Journal of Speech-Language Pathology, 23*(1), 73–84. https://doi.org/10.1044/1058-0360(2013/12-0164

Thompson Burdine, J., Thorne, S., & Sandhu, G. (2021). Interpretive description: A flexible qualitative methodology for medical education research. *Medical Education, 55*(3), 336-343.

Thompson, R. J., O'Quinn, A. N. & Logue, P. E. (1979). Gilles de la Tourette syndrome: a review and neuropsychological aspects of four cases. *Journal of Pediatric Psychology, 74*(4), 371–387. https:// doi.org/10.1177/014107688607900508

Thorne, S. (2016). *Interpretive description*. Left Coast Press.

Toppelberg, C. O., & Shapiro, T. (2000). Language disorders: A 10-year research update review. *Journal of the American Academy of Child and Adolescent Psychiatry, 39*(2), 143–152. https://doi.org/10.1097/00004583-200002000-00011

Tourette Association of America. (2021). *Homeschooling: A flexible, fluid, child-focused education.* https://tourette.org/homeschool/

Towbin, K. E., Pradella, A., Gorrindo, T., Pine, D. S., & Leibenluft, E. (2005). Autism spectrum traits in children with mood and anxiety disorders. *Journal of Child and Adolescent Psychopharmacology, 15*(3), 452–464. https://doi.org/10.1089/cap.2005.15.452

Travis, E., & Juarez-Paz, A. V. O. (2020). Experiences of Tourette syndrome caregivers with supportive communication. *Qualitative Health Research, 30*(10), 1480–1490. https://doi.org/10.1177/1049732320915444

Tricco, A. C., Lillie, E., Zarin, W., O'Brien, K. K., Colquhoun, H., Levac, D., Moher, D., Peters, M. D. J., Horsley, T., Weeks, L., Hempel, S., Akl, E. A., Chang, C., McGowan, J., Stewart, L., Hartling, L., Aldcroft, A., Wilson, M.G., Garritty & Straus, S. E. (2018). PRISMA extension for scoping reviews (PRISMA-ScR): checklist and explanation. *Annals of Internal Medicine, 169*(7), 467–473. https://doi.org/10.7326/M18-0850

United States Census Bureau (2022a). *Census Bureau releases new educational attainment data*. https://www.census.gov/newsroom/press-releases/2022/educational-attainment.html

United States Census Bureau (2022b). *What languages do we speak in the United States?* https://www.census.gov/library/stories/2022/12/languages-we-speak-in-united-states.html

United States Census Bureau (2023) *Quick facts*. https://www.census.gov/quickfacts/fact/table/US/PST045223

Vaïsänen, R., Loukusa, S., Moilanen, I., & Yliherva, A. (2014). Language and pragmatic profile in children with ADHD measured by children's communication checklist 2nd edition. *Logopedics Phoniatrics Vocology, 39*(4), 179–187. https://doi.org/10.3109/14015439.2013.784802

Van Steensel, F. J. A., Bögels, S. M., & Wood, J. J. (2013). Autism spectrum traits in children with anxiety disorders. *Journal of Autism and Developmental Disorders, 43*(2), 361–370. https://doi.org/10.1007/s10803-012-1575-z

Van Widenfelt, B. M., Goedhart, A. W., Treffers, P. D. A., & Goodman, R. (2003). Dutch version of the Strengths and Difficulties Questionnaire (SDQ). *European Child and Adolescent Psychiatry, 12*, 281–289.

Verté, S., Geurts, H. M., Roeyers, H., Oosterlaan, J., & Sergeant, J. A. (2005). Executive functioning in children with autism and Tourette syndrome. *Development and Psychopathology, 17*(2), 415–445. https://doi.org/10.1017/S0954579405050200

Wadman, R., Glazebrook, C., Beer, C. & Jackson, G. M. (2016). Difficulties experienced by young people with Tourette syndrome in secondary school: A mixed methods description of self, parent and staff perspectives. *BMC Psychiatry, 16*(1), 1–11. https://doi.org/10.1186/s12888-016-0717-9

Walenski, M., Mostofsky, S. H., & Ullman, M. T. (2007b). Speeded processing of grammar and tool knowledge in Tourette's syndrome. *Neuropsychologia, 45*(11), 2447–2460. https://doi.org/10.1016/j.neuropsychologia.2007.04.001.

Wiig, E. H., & Secord, W. A. (1989). *Test of Language Competence (TLC).* Merill.

Wiig, E. H., Semel, E., & Secord, W. A. (2013). *Clinical Evaluation of Language Fundamentals (CELF-5)* (5th ed.). Pearson.

Wiig, E. H., Semel, E., & Secord, W. A. (2013). *Clinical Evaluation of Language Fundamentals Screening Test* (5th ed.). Pearson.

Williams, M. T., Mugno, B., Franklin, M., & Faber, S. (2014). Symptom dimensions in Obsessive-Compulsive Disorder: Phenomenology and treatment outcomes with exposure

and ritual prevention. *Psychopathology, 46*(6), 365–376. https://doi.org/10.1159/000348582.

Wilson. K, & Prizant, B. (2022). *Turning kids' passions and enthusiasms into superpowers. In Diverse thinking different learning* [Podcast]. Retrieved from https://childnexuspodcast.com/ep-80-turning-kids-passions-and-enthusiasms-into-superpowers-with-barry-prizant-ph-d-ccc-slp/

Wiseman-Hakes, C., Kakonge, L., Doherty, M., & Beauchamp, M. (2020). A conceptual framework of social communication: Clinical applications to pediatric traumatic brain injury. *Seminars in Speech and Language, 41*(2), 143-160.

Woerner, W., Becker, A., & Rothenberger, A. (2004). Normative data and scale properties of the German parent SDQ. *European Child and Adolescent Psychiatry, 13*(2), 3–10.

Wu, M. S., & McGuire, J. F. (2018). Psychoeducation about tic disorders and treatment. In J. F. McGuire, T. K. Murphy, J. Piacentini, & E. A. Storch (Eds.), *The clinician's guide to treatment and management of youth with Tourette syndrome and tic disorder* (pp. 21–41). Elsevier Academic Press. https://doi.org/10.1016/B978-0-12-811980-8.00002-9

Yeates, K. O. & Bornstein, R. A. (1994). Attention deficit disorder and neuropsychological functioning in children with Tourette's syndrome. *Neuropsychology, 8*, 65–74. https://doi.org/10.1037/0894- 4105.8.1.65

Yew, S. G. K., & O'Kearney, R. (2013). Emotional and behavioural outcomes later in childhood and adolescence for children with specific language impairments: Meta-analyses of controlled prospective studies. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 54*(5), 516–524. https://doi.org/10.1111/jcpp.12009

Zhou & Brown (2015). *Educational learning theories* (2nd ed.). Education Open Textbooks.

Zhu, Y., Leung, K. M., Liu, P. Z., Zhou, M., & Su, L. Y. (2006). Comorbid behavioural problems in Tourette's syndrome are positively correlated with the severity of tic symptoms. *Australian and New Zealand Journal of Psychiatry, 40*(1), 67–73. https://doi.org/10.1111/ j.1440-1614.2006.01745.x

Appendix A

Search Strategy and Dates Searched

Database: OVID MEDLINE (1946-present) Date: November 7, 2019 Searched by: Angela Feehan Number of results: 270 Search strategy: 1. (Language skills or language characteristics or high level language impairment or discourse impairment or normal communicative performance or language profiles or 'concreteness of language' or poor language formulation or frontal cortex language involvement or language development or language disorders or grammar or language or linguistic knowledge or idiosyncratic knowledge or mental lexicon or regular past tense or irregular past tense or linguistics or language tests or learning disabilities or developmental dyslexia or dyslexia or learning disorders or social decision making or social communication or social communication deficits or social communication questionnaire or sarcastic remarks or mentalising or pragmatic language or sarcasm or 'theory of mind' or social cognition).mp.[mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms] 2. limit 1 to English language 3. Child Language/ or Language Development/ or Language/ or Communication Disorders/ or Language Disorders/ or Language Development Disorders/ or Speech Language Pathology/ or Semantics/ or Vocabulary/ or Metaphor/ or Narration/ or Social Communication Disorder/ or 'Theory of Mind'/ or Social Skills/ or Nonverbal Communication/ or Facial Expression/ or Social Perception/ or Interpersonal Relations/ or Literacy/ or Writing/ or Reading/ or Agraphia/ or Dyslexia/ or Learning Disorders/ or Language Tests/ or 'Rehabilitation of Speech and Language'/ or Language Therapy/ 4. limit 3 to English language 5. 2 or 4 6. Tourette Syndrome/ or

tourette*.mp. 7. limit 6 to English language 8. 5 and 7 The following databased were searched on February 9th, 2022: OVID EMBASE (1974-present) EBSCOHost CINAHL Plus with full text (1937-present) EBSCOHost ERIC EBSCOHost Education Research Complete EBSCOHost PsycINFO Bielefeld Academic Search Engine (BASE): ProQuest Dissertations & Theses

Appendix B

Scoping Review Studies

	Lead author	Year	Study type	Participant groups	Skills measured
1	Baron-Cohen	1995	Descriptive	TS ASD TS+ASD	Theory of mind
2	Bawden	1998	Observational	TS Diabetes Mellitus	Social interaction
3	Bornstein	1990	Descriptive	TS	Verbal intelligence
4	Bornstein	1983	Descriptive	TS	Receptive vocabulary Verbal intelligence
5	Bornstein	1991	Descriptive	TS	Verbal intelligence
6	Brand	2002	Observational	TS TS+ADHD	Verbal fluency
7	Brookshire	1994	Observational	TS Normal Siblings Arithmetic Disability	Verbal fluency
8	Carter	2000	Observational	TS TS+ADHD Nondisabled peers	Verbal intelligence Social interaction
9	Channon	2003	Observational	TS TS+ADHD TS+OCD Nondisabled peers	Verbal fluency Story recall
10	Church	2009	Observational	TS Nondisabled peers	Verbal fluency
11	Claussen	2018	Descriptive	TS	Prevalence of language problems
12	Comings	1991	Observational	TS with an autistic relative ASD+TS	Language development in cases
13	Cravedi	2018	Descriptive	Pure TS Complex TS TS+Attention & writing problems	Verbal intelligence Expressive and receptive language

	Lead author	Year	Study type	Participant groups	Skills measured
14	Darrow	2017	Observational	TS TS+ADHD TS+OCD TS+ADHD+OCD Family members of people with TS	Social communication Social interaction Prevalence of autistic traits
15	De Groot	1997	Observational	TS TS+ADHD TS+OCD TS+ADHD+OCD	Verbal intelligence Verbal fluency Categorization
16	Drury	2018	Observational	TS TS+ADHD Nondisabled peers	Sarcasm comprehension Verbal fluency (Non)mentalistic questions
17	Drury	2012	Observational	TS TS+ADHD Nondisabled peers	Emotion perception
18	Dye	2016	Observational	TS Nondisabled peers	Non-word repetition
19	Dykens	1990	Observational	TS TS+ADHD	Social communication
20	Eapen	2019	Observational	TS ASD Nondisabled peers	Social communication Social interaction Autistic traits
21	Ferrari	1984	Descriptive	TS	Receptive vocabulary Verbal intelligence
22	Gallina	1989	Observational	TS ADHD Nondisabled peers	Social interaction
23	Gorman	2010	Observational	TS Nondisabled peers	Social interaction
24	Güler	2015	Observational	TS Nondisabled peers	Social communication Social interaction Social cognition Autistic traits
25	Hagin	1988	Descriptive	TS	Verbal intelligence
26	Harris	1995	Observational	TS ADHD	Verbal fluency

	Lead author	Year	Study type	Participant groups	Skills measured
				TS+ADHD	
27	Hoekstra	2004	Descriptive	TS	Social interaction
28	Huckeba	2008	Observational	TS Nondisabled peers	Verbal intelligence
29	Hulbert	1986	Descriptive	TS	Social interaction Naming Sentence repetition Following directions Verbal intelligence Verbal fluency
30	Incagnoli	1983	Descriptive	TS	Verbal intelligence
31	Jensen	2004	Observational	TS Dyslexia Tics+Dyslexia	Verbal fluency Receptive vocabulary Expressive vocabulary
32	Kadesjö	2000	Descriptive	TS	Social interaction Autistic traits
33	Kalafayan Sweeten	1997	Descriptive	TS	Receptive vocabulary verbal intelligence Language development in a case
34	Khalifa	2010	Observational	TS Nondisabled peers	Verbal intelligence Verbal fluency
35	Lanser	1993	Observational	TS Right hemisphere dysfunction	Verbal intelligence
36	Legg	2005	Descriptive	TS Nondisabled peers	Higher-level language Narrative language
37	Ludlow	1982	Observational	TS Nondisabled peers	Language expression Language comprehension Sentence repetition
38	Mahone	2002	Observational	TS ADHD TS+ADHD Nondisabled peers	Verbal fluency
39	Mahone	2001	Observational	TS ADHD Nondisabled peers	Verbal fluency

	Lead author	Year	Study type	Participant groups	Skills measured
40	Marek	2006	Descriptive	TS	Social interaction
41	O'Quinn	1980	Descriptive	TS	Language development in cases Verbal intelligence
42	Pratt	2000	Observational	TS Nondisabled peers	Story recall Naming
43	Rajendran	2005	Observational	TS ASD (Asperger syndrome) Nondisabled peers	Social cognition (sarcasm, figures of speech, inappropriate requests)
44	Schuerholz	1996	Observational	TS TS+ADHD TS+QueryADHD Unaffected siblings	Naming Verbal fluency
45	Schuerholz	1998	Observational	TS TS+ADHD ADHD Nondisabled peers	Naming Verbal fluency
46	Shapiro	1988	Observational	TS TS+ADHD Psychiatric referral Nondisabled peers	Verbal intelligence
47	Shapiro	1974	Descriptive	TS	Verbal intelligence
48	Spencer	1998	Observational	TS TS+ADHD ADHD Psychiatric referral Nondisabled peers	Prevalence of language disorder
49	Stokes	1991	Descriptive	TS	Receptive vocabulary Verbal intelligence
50	Sukhodolsky	2003	Observational	TS ADHD TS+ADHD Nondisabled peers	Social communication Social interaction
51	Takács	2018	Observational	TS Nondisabled peers	Story recall
52	Thompson	1979	Descriptive	TS	Verbal intelligence

	Lead author	Year	Study type	Participant groups	Skills measured
					Language development in cases
53	Verté	2005	Observational	TS ASD (high functioning) TS+ASD (high functioning) Nondisabled peers	Verbal fluency Categorization Social communication Autistic traits
54	Wadman	2016	Descriptive	TS	Autistic traits
55	Walenski	2007	Observational	TS Nondisabled peers	Past tense production Naming
56	Yeates	1994	Observational	TS TS+ADHD	Verbal intelligence

Appendix C

Survey Background Questions

Please answer the following background questions about your child:

Background questions, Part 1/5

Who diagnosed your child with Tourette syndrome?

	Family doctor	Pediatrician	Psychiatrist	Neurologist	Other
	○	○	○	○	○

At about what age was your child diagnosed with Tourette syndrome? _____

	8	8.5	9	9.5	10	10.5	11	11.5	12	12.5	13	13.5	14	14.5	15	15.5	16	16.5
What is your child's current age?	○	○	○	○	○	○	○	○	○	○	○	○	○	○	○	○	○	○

Background questions, Part 2/5

	Male	Female
What is your child's sex? (sex assigned at birth)	○	○

	Girl	Boy	Trans	Not listed
What is your child's gender?	○	○	○	○

Where are you located?

○ United States
○ Canada

Background questions, Part 3/5

Please indicate if your child has any of the following diagnoses:

	Yes	No	Suspected
Attention deficit hyperactivity disorder (ADHD)	○	○	○
Obsessive Compulsive Disorder (OCD)	○	○	○
Obsessive compulsive behaviours (but not OCD)	○	○	○
Learning disability (LD)	○	○	○
Anxiety disorder	○	○	○
Depression	○	○	○
Other mood disorder	○	○	○
Hearing loss	○	○	○
Language/Communication disorder	○	○	○
Speech disorder	○	○	○

Please list any other serious medication conditions not listed above that your child has (e.g., cancer, epilepsy, cerebral palsy): _____

Background questions, Part 4/5

What language do you speak most of the time in your home? (e.g., French) _____

	10%	20%	30%	40%	50%	60%	70%	80%	90%	100%
What percentage of the time do you speak the language you listed above?	○	○	○	○	○	○	○	○	○	○

Do you speak any other languages in your home? ○ Yes ○ No

	I did not complete high school	I completed high school (or equivalent)	I have a college degree (or similar)	I have a bachelor's degree	I have a graduate degree
What is your level of education?	○	○	○	○	○

Background questions, Part 5/5

What racial or ethnic group does your child belong to?

Arab	Black	Chinese	Filipino	Indigenous	Japanese	Korean	Latin American	South Asian	South-east Asian	West Asian	White	Not listed
☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

Thank you for answering our background questions.

Next, you will answer questions about your child's communication.

Appendix D

Parent Communication Log Book Example

Day 1 Positive Example

Think of a situation where communication went ***well*** today. It can be any interaction that your child was part of. Briefly describe it.

What do you think was happening that may have affected this situation?

What did the other person/people do in the situation?

Page Break

Day 1 Negative Example

Think of a situation where there were ***problems*** in communication today. It can be any interaction that your child was part of. Briefly describe it.

What do you think was happening that may have affected this situation?

What did the other person/people do in the situation?

Page Break

Appendix E

Interview Guide

1. I'm hoping this interview will help me understand CHILD's communication skills. When I say communication, I'm talking about how CHILD gets his message across to others in his day-to-day life and how he/she understands what others are trying to say to him/her. Do you have any general thoughts about how CHILD communicates?

Notes on theory: this question establishes my theoretical lens on communication as "functional"

2. What are the activities that your child does besides spending time at school and home? How do they do with interacting with others in <each setting>?

Note on theory: Get an idea of what everyday contexts the child communicates in and ask about each one. This will help me to cover the "microstructure"

3. I'm interested in hearing some stories about how CHILD communicates at home. Can you tell me about X situation from your 7-day log book? You wrote that X happened (summarize what is written in the log to remind parent). Can you please tell me the story of what happened?

Notes on theory: try to get elaboration form the functional examples in the log book will enable me to recreate functional examples of communication.

4. How does CHILD do with getting his/her message across with other kids? Siblings? Peers? Does CHILD get along well with other kids? Does CHILD enjoy the company of other kids? What helps him/her? What makes things harder?

Notes on theory: this question asks about "individual" and the "microstructure" levels of Bronfenbrenner's bioecological model, while focusing on examples of functional communication (functionalism)

5. What does CHILD find easy/difficult about being at school. What subjects does CHILD like/dislike? What do teachers say about CHILD's ability to follow along with what's happening in the classroom? Now? In the past? What helps CHILD in school? What makes things harder?

Notes on theory: Asks about school as part of the child's "microstructure." Tries to bring out supports and challenges.

6. When CHILD wants to tell you about something that happened and you weren't there, is he/she able to explain the situation to you clearly? Do you do/have you done anything to help him/her with giving these explanations? Please tell me about a time that happened.

Notes on theory: Asks about a more complex contextual communication situation, inviting the parent to talk about the interaction of different factors (e.g., their behaviour in the situation). Tries to get functional communication examples. Looks at supportive factors.

7. Is CHILD able to participate in conversations at home, understanding what's being said and saying things that add to the conversation? What do you notice about the way CHILD communicates?

Notes on theory: Ask about the home environment communication context as part of the "microstructure"

8. Is there anything you'd like to add about what's already been said? Is there anything else about CHILD's communication that I didn't ask you about?