PROFESSIONAL DOCTORATES

Developing Systemic Approaches to Tic Disorders in Education and Healthcare Settings

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Kramer, Jaxon

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Developing Systemic Approaches to Tic Disorders in Education and Healthcare Settings

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North Wales Clinical Psychology Programme

Bangor University

A thesis submitted in partial fulfilment for the degree of Doctorate in Clinical Psychology

June 2023

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Declaration

I hereby declare that this thesis is the results of my own investigations, except where otherwise stated. All other sources are acknowledged by bibliographic references. This work has not previously been accepted in substance for any degree and is not being concurrently submitted in candidature for any degree unless, as agreed by the University, for approved dual awards.

Yr wyf drwy hyn yn datgan mai canlyniad fy ymchwil fy hun yw'r thesis hwn, ac eithrio lle nodir yn wahanol. Caiff ffynonellau eraill eu cydnabod gan droednodiadau yn rhoi cyfeiriadau eglur. Nid yw sylwedd y gwaith hwn wedi cael ei dderbyn o'r blaen ar gyfer unrhyw radd, ac nid yw'n cael ei gyflwyno ar yr un pryd mewn ymgeisiaeth am unrhyw radd oni bai ei fod, fel y cytunwyd gan y Brifysgol, am gymwysterau deuol cymeradwy.

Jaxon Kramer

Date: 30/05/2023

Acknowledgements

"Children are the living messages we send to a time we will not see."

— Neil Postman

I would like to begin my expressing my profound gratitude to my supervisor and training coordinator Dr Mike Jackson. Your enduring support and guidance brought me solace and encouragement when I needed it the most. So, "Old Salt" I say thank you, and wish you fair winds and following seas. I would also like to extend my sincere thanks to Dr Chris Saville, Dr Maddie Groom and Dr Sophie Hall, your contributions and continued counsel throughout this research project was invaluable. To Dr Rachel Skippon, who's patience and nurturing during this project cannot be overestimated. Importantly, to the healthcare experts who sacrificed their free time to participate in the Delphi study, thank you for your generosity, expertise, and kindness; without you this project would not have been possible.

To my Father, your guiding hand, unconditional love, and endless affection helped me embrace my Tourettes. You showed me what it means to be "Tourettian," and I wouldn't be where I am without you. To my brothers Jeremy and Jordan, your love and friendship has been a cornerstone of my life, and I am better because of you. Thank you to my sister-in-law Molly, who's advice and reflective conversations I cherish, and my niece Moss who reminds me how important it is to be silly and laugh often.

To my fellow trainees in the great Covid cohort of 2020, thank you for being such a sturdy crew…*Relentlesssss.* To Dr Katie Bigham, thank you for improving my life with your advice, friendship, shared laughter, and boundless compassion. Finally, to my fellow "Tourettians" across the globe: You are remarkable and courageous, *keep going*.

Dedication

This thesis is dedicated to my mother Vikki Kramer who gave me the everlasting gift of knowing what it feels like to be loved absolutely, even when I didn't see a reason why. She dedicated her life's work to improving the lives of children with Tourette Syndrome around the globe. My mother truly was a champion of children. No matter how old I get I am, and always will be, your *elskan mín*.

Thesis Abstract

In chapter one, a scoping review and narrative synthesis of empirical evidence from nine studies reporting primary data related to school-based support strategies for children and young people (CYP) with chronic Tic Disorders (TD) was carried out. Findings revealed a paucity of empirical evidence and significant heterogeneity among included studies. Four thematic sub-groups were used to discuss key findings: limited knowledge and experience of teachers and school-staff; learning: barriers, accommodations, and classroom strategies; enhancing understanding and empathy in peers of CYP with TD; and effective systemic communication.

In Chapter two, an empirical study aimed to build consensus among healthcare experts to: define and characterise a realistic service model of care with a remit of assessing, diagnosing, and treating TD in CYP; and to identify potential obstacles and facilitators to establishing and implementing said service model. A panel of ten experts participated in a three-round e-Delphi study. In Round one, experts provided free-text responses to 7 open-ended questions. Researchers qualitatively generated 28 statements from round one responses which experts rated on relative agreement and/or importance in subsequent survey iterations across Round two and three. The study was successful in gaining expert consensus on key aspects comprising the proposed model of care for CYP with TD: service configuration, funding arrangements, operational structure, interventions and barriers and facilitators to service implementation. A ranking hierarchy identifying prioritised professional roles for newly commissioned services was also created.

In Chapter three, the implications of the findings from the first two papers are collectively considered in the context of theory development, clinical practice and future research. The thesis then concludes with a reflective commentary from the first author.

Chapter One

Scoping Review and Narrative Synthesis

School-Based Support for Children and Young People with Chronic Tic Disorders: A Scoping Review

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Abstract

Children and Young People (CYP) with chronic Tic Disorders (TD) are estimated to account for 3% of mainstream students worldwide. The school experiences of CYP with TD are often negative, ranging from TD-related learning difficulties, to unhelpful responses from teachers and victimisation from peers. TD have been associated with poor quality of life, impaired social-emotional development, and other adverse long-term outcomes. Therefore, creating inclusive school environments that are sensitive to the needs of CYP with TD is crucial. This study aimed to review existing empirical evidence relating to school-based support for CYP with TD. A scoping review and narrative synthesis of empirical evidence was carried out using literature identified through a comprehensive search of four databases (ProQuest, Embase, Scopus, ESBCO), and subsequent searches for grey literature and citation-chaining. Systematic screening resulted in a total of nine studies being included in the present review. Findings revealed a paucity of empirical evidence related to providing school-based support for CYP with TD. There was significant heterogeneity among included studies. Four thematic sub-groups were used to discuss key findings: Limited Knowledge and Experience of Teachers and School-Staff, Learning: Barriers, Accommodations, and Classroom Strategies, Enhancing Understanding and Empathy in Peers of CYP with TD, and Effective Systemic Communication. This review provides an important mapping of empirical evidence and digest of their respective key findings related to school-based support for CYP with TD. Implications of key findings for clinical practice and future research is discussed.

Keywords: Tic Disorders, Tourette, School, School-based, Support, Children

Introduction

Overview of Tic Disorders: Clinical Characteristics, Prevalence and Comorbidities

Chronic Tic Disorders (TD) are polygenic inherited neuro-developmental conditions (Abdulkadir et al., 2022), characterised by the presence of motor and/or vocal tics. Tics are defined in the Diagnostic and Statistical Manual, fifth edition (DSM-5; American Psychiatric Association [APA], 2013) as "sudden, rapid, recurrent, but non-rhythmical motor movements and/or vocalizations, generally preceded by premonitory somatosensory urges." The age of onset for TD is typically between the ages of six and seven years-old (Gloor & Walitza, 2016), with tics waxing and waning in frequency, intensity, variety, and complexity. Tic severity in Children and Young People (CYP) with TD often peaks in puberty (Black et al., 2020), and demonstrate an ability to suppress tic expression for short periods of time, however patients report an intense increase in symptoms shortly thereafter (Ueda & Black, 2021). The DSM-5 outlines three main categories of TDs:

1. Provisional Tic Disorder
2. Persistent (chronic) Motor or Vocal Tic Disorder
3. Tourette Syndrome (TS; also known as Tourette Disorder)

CYP with TD are estimated to account for around 3% of all children in mainstream schools worldwide (Adams et al., 2023), and 1% of school-aged children in the United Kingdom (UK; Hall et al., 2022). This suggests TD to be more common than previously believed, with prevalence rates that nearly parallel those of Autism Spectrum Condition in school-aged children in the UK (Cavanna et al., 2020). TD are shown to affect biological males more than biological females, with a male to female preponderance ranging between 3:1 and 4.3:1 (Szejko et al., 2022). International studies have also established TD to be highly comorbid with other neuropsychiatric presentations, with the most common being Attention Deficit-Hyperactivity Disorder (ADHD) occurring in between 50% to 80% of patients with

TD, and Obsessive-Compulsive Disorder (OCD) in 20% to 60% respectively (Pedersen et al., 2022). In research, presentations of TS/TD with comorbid conditions such as ADHD and/or OCD are commonly referred to as TS+ (Müller-Vahl et al., 2022). Additionally, in the literature base, referring to the term TS is often to include other forms of TD (Szejko et al., 2022). However, with the intention of embracing inclusivity, the present review chose an opposite approach, adopting the term TD to encompass all chronic tic disorders, including TS (unless referenced to specifically).

Impact of TD on CYP

TD has been associated with significantly poorer Quality of Life (QoL) in CYP compared to the respective typically developing population, with substantial evidence demonstrating the marked distress and impairment CYP with TD experience across multiple life domains (Evans et al., 2016). TD in CYP have been associated with adverse physical health outcomes such as: sleep disorders occurring in 64% of paediatric patients with TD (Hibberd et al., 2020), neuropathic pain, and physical injury (e.g. bone fractures and tissue damage) as a consequence of tic expression (Uzun Cicek et al., 2020; Fusco et al., 2006). Academically, CYP with TD often struggle with: concentrating or attending to oration from teachers, performing well on time-oriented tasks, writing, and commonly avoid reading aloud, asking questions, and completing homework (Claussen et al., 2018). These difficulties are also further amplified by common comorbidities such as ADHD and OCD (Lowe et al., 2019). Furthermore, evidence reveals that teachers are often reported to respond and/or view tic expression as disruptive and/or attention-seeking behaviour; often resulting in CYP with TD being criticised or disciplined in front of the class, secluded, or even ejected from the classroom as punishment (Set & Warner, 2021).

The negative impact of TD on CYP's psychological and social-emotional functioning is well evidenced. CYP with TD often report feelings of shame, hopelessness, and intense

guilt in relation to perceptions of being a burden on caregivers and/or loved ones (Edwards et al., 2017; Happich, 2012). TD is also associated with poor self-concept (how one views or thinks of themselves) and self-esteem (one's subjective experience of their self-concept). In qualitative studies, CYP with TD express feelings of self-hatred; describing themselves as freaks, werewolves, or aliens incapable of integrating into the world around them (Rindner, 2004; Lee et al., 2019). Importantly, these negative self-experiences are often socially-constructed through the internalisation of the stigmatisation, victimisation and bullying CYP with TD frequently face from peers (Lee et al., 2016; Edwards et al., 2017; Kim & Tak, 2020). School is a setting which is fundamental to interrelated areas of development, and the negative experiences CYP with TD face in this setting has been shown to have long-lasting adverse effects which they carry into adulthood (Malli & Forrester-Jones, 2022). Currently, there are no known school-based support programmes or interventions in the UK that are tailored to supporting CYP with TD.

Current Approaches to School Support in UK

As outlined in the Department for Education's (DfE) statutory guidance on Special Education Needs (SEN; 2020), all mainstream primary and secondary schools in the UK are required to have a Special Educational Needs Coordinator (SENCo). CYP are reported as having SEN when "they have a learning difficulty or disability which calls for special educational provision to be made for him or her" (DfE, 2020, p.15). The role of SENCo is described in the SEN guidance as having "day-to-day responsibility for the operation of SEN policy and coordination of specific provision made to support individual pupils with SEN" (DfE, 2020, p.108). To become a SENCo, certified teachers must undergo further post-graduate training which has been reported to vary greatly in regards to topics covered in curriculum (Esposito & Carroll, 2019). SENCos play an important role in establishing inclusive learning environments for CYP with SEN (Cole, 2005). However, research has

consistently highlighted the disparity in how the SENCo is operationalised in government policies and the reality of how the role is interpreted and applied in practice (Dobson, 2019). Factors such as: the growing number of SEN students, insufficient specialised training, clarity of role responsibilities, lack of support and the solitary nature of the SENCo role have been evidenced as impacting the facilitation of school-based support for CYP (Beaton et al., 2021).

Rationale and Objectives

Evidence has established that TD in CYP is not only more common than once believed, but the adversities they face, particularly in school settings (key environments for development) have a significant impact on their overall QoL, learning, and social-emotional development. Therefore, many mainstream teachers and school staff will likely be confronted with educating CYP with TD, and appropriately managing these presentations is critical for their well-being. Additionally, improving our understanding of supporting CYP with TD in school-settings is necessary in the context of inclusivity and the UK's national (DfE, 2020) and international (United Nations, 2016) legal requirements and commitments. The growing evidence on the effectiveness of various clinical interventions for TD in CYP has been widely reviewed. However, to the authors' knowledge, there are no reviews focusing on empirical evidence reporting primary data on school-based support strategies for CYP with TD. Therefore, this paper aims to perform a scoping review and narrative synthesis of the existing empirical evidence relating to school-based support for CYP with TD. Additionally, as recommended by the Joanna Briggs Institute (JBI; Peters et al., 2020), the present scoping review was guided by the following research questions which were conceptualised using the PCC framework (Table 1):

1. What are the characteristics of the empirical studies?
2. What does the empirical evidence reveal about strategies to support CYP with TD in school settings, and what are the gaps in current knowledge?

Table 1

PCC Framework Elements and Present Scoping Review's Definitions

PCC Element	Definition
Population	CYP with TD
Concept	Support Strategies
Context	School-based Settings

Methods

A scoping review was the employed method of addressing the aforementioned research questions as it is a useful approach to identifying and mapping types of available evidence in a particular field, and ascertaining relative gaps in knowledge (Munn et al., 2018). The present scoping review was conducted in accordance with guidance from the JBI (2020) and the Preferred Reporting Items for Scoping Reviews framework (PRISMA-ScR; Tricco et al., 2018).

Eligibility

Inclusion and Exclusion criteria (Table 2) informed by the PCC framework were established a priori to determine the eligibility of publications for the present review. Due to the limited availability of empirical research on school-based support for CYP with TD, no limitation was placed on year of publication, and studies employing a range of research designs were included, such as: qualitative, cross-sectional, mixed-methods, and case designs. Publications were only included if they: had Tourette Syndrome or Tic Disorders in

the title, were written in the English language, reported on primary data specifically pertaining to support for CYP (18 years and under) with TS and/or TD in school settings up to and including Further Education (i.e., GCSE or A-Level). Due to the high rates of comorbid conditions shown to occur in CYP with TD, studies reporting primary data CYP with TD and co-morbid presentations (e.g., ADHD, OCD) were included as long as school-based support for TD was the substantial focal point for discussion in the paper. Publication types such as reviews, opinions, and protocols were excluded as they would not report on primary data. Additionally, to improve the efficiency of the search strategy, studies with prevalence or incidence in title were excluded, as these papers would address epidemiological findings rather than school-based support.

Table 2

Eligibility Criteria

Inclusion Criteria	Exclusion Criteria
Publication has Tourette Syndrome or Tic Disorders in Title	Publication written in any language other than English
Reports primary data on supporting children and young people 18 years and under.	Reports primary data focusing on school-settings above Further Education Level (e.g., University/Higher Education).
Reports primary data specifically pertaining to support for Tourette and/or Tic Disorders in school settings up to and including Further Education such as i.e., GCSE or A-Level).	Reports primary data on supporting populations over the age of 18.
Reports primary data in which supporting Tourette Syndrome and/or Tic Disorders is substantially focused/discussed in the publication, co-morbid presentations may be included in the data if these co-morbidities are not given primacy over TS and/or TD in terms of focus of study/discussion of support.	Reports primary data in which co-morbid presentations are the primary focal point for support.
	Does not report primary data
	Publication has prevalence or incidence in title

Search Strategy and Publication Selection

A comprehensive search strategy was developed and refined through research team discussion. A systematic search of four databases (ProQuest, Embase, Scopus, ESBCO) was conducted. Subsequent searches included grey literature and citation chain-searching, and the final literature search was carried out in April 2023 using the search terms and script displayed in Table 3.

Table 3

Search Terms with Boolean Operators used in Database Searches

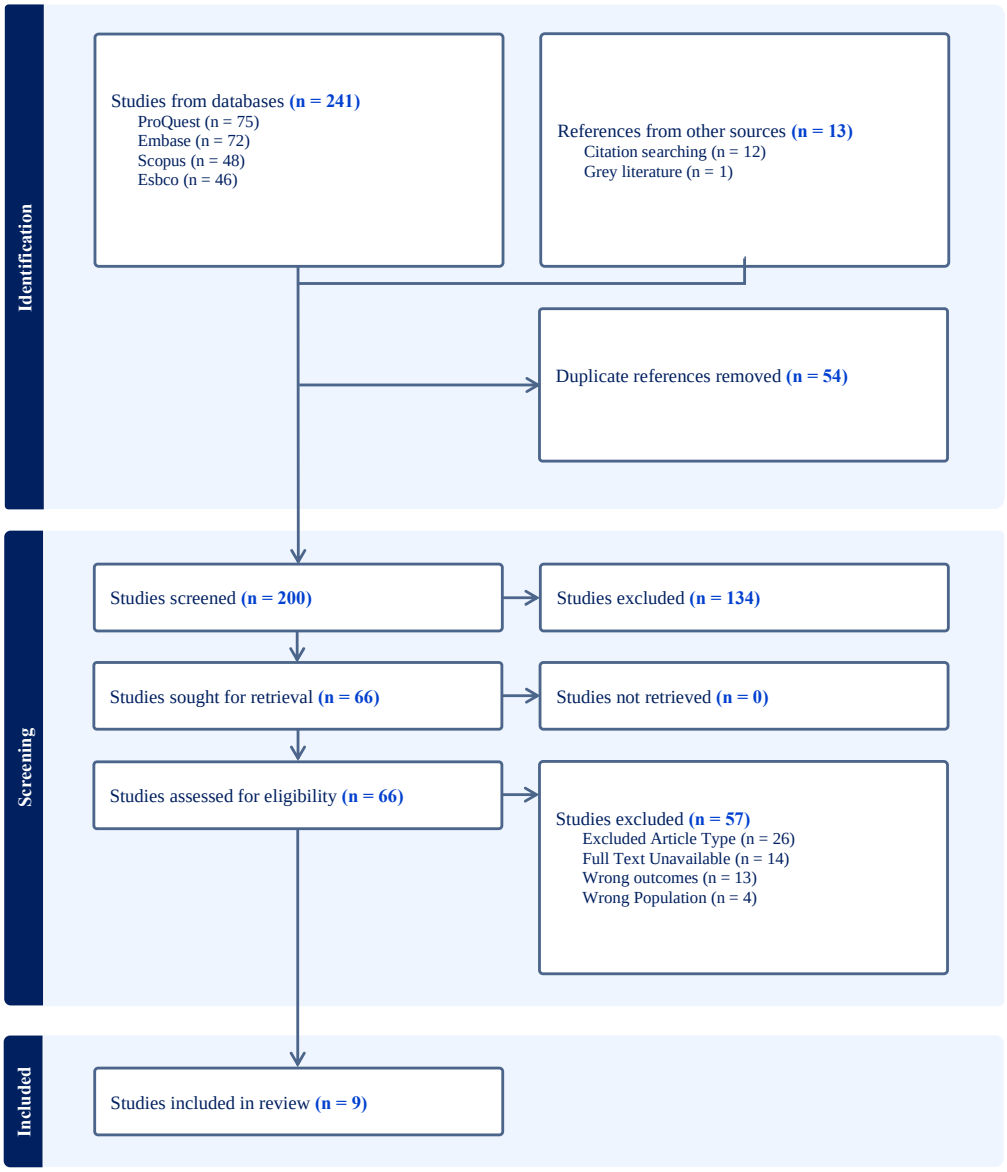
Search terms
Tourette* OR "tic disorder" OR "tic disorders" OR Tics [limited to title]
AND
school* OR school-based OR teacher* OR school AND setting
[Limited to Title/Abstract/Keyword]
AND NOT
prevalence OR incidence
[limited to title]

As illustrated in the PRISMA Diagram (Figure 1) 254 references were imported to Zotero reference management software and subsequently exported to Covidence software. Initially, 54 duplicate references were removed, and 200 studies were screened against title and abstract resulting in 134 studies being excluded. Subsequent full-text eligibility assessments were then carried on the remaining 66 studies, resulting in 57 studies being excluded based on: excluded article Type (n=26); full text being unavailable (n=14); wrong outcomes being measured (n=13); and wrong population (n=4). The titles and abstracts of a

randomly selected sample of papers (n=20) were screened against eligibility criteria (Table 2) by both first and second authors to ensure inter-reviewer reliability (IRR). Authors were in agreement on 19 of the 20 papers (IRR = 95%), with the single discrepancy being resolved through discussion between authors. Finally, nine studies were considered eligible following assessment and included in this review.

Fig. 1

PRISMA Diagram Illustrating Search Strategy and Study Selection



Data Extraction and Charting Process

Based on the present scoping review's research topic and guidance from the JBI (Peters et al., 2020), a standardised form (Appendix A) was developed and employed in the data extraction and charting process. The key features and information of each included study were extracted and charted using the standardised forms.

Narrative Synthesis

The present scoping review methodology employed a comprehensive and systematic process to the: identification, screening and inclusion of studies, and the extraction and charting of their data (Tricco et al., 2018). Due to the heterogeneity of included articles (i.e. research design and data outcomes), the amalgamation and discussion of key findings from included studies was achieved through a modified three stage format of narrative synthesis, informed by guidance from Popay et al. (2006) and outlined in Table 4.

Table 4

Stages and Processes Involved in the Modified Format of Narrative Synthesis

Stage	Process
1. Developing the preliminary synthesis	Data extraction and charting
2. Comparing themes within and between studies	Extracted data analysed to uncover emerging themes in the findings from each study. This allowed for key findings from heterogenous studies to be synthesised into homogenous groups for further discussion.
3. Reporting/Discussion of key findings based on thematic grouping	Thematic sub-groups were then used to discuss key findings from included studies in the context of the overarching theme which reflected the primary research question: *School-Based Support for CYP with TD*

Results

Table 5 summarises the key characteristics of the nine studies included in the present scoping review. Year of publications ranged from 2005 to 2022, eight out of the nine studies were published peer-reviewed articles; the ninth being a doctoral dissertation (Fine, 2020).

Design

Of the nine included studies: three were qualitative (Fine, 2020; Grace & Russell, 2005; Ludlow et al., 2022), three adopted varying forms of experimental designs (Gilman et al., 2005; Holtz & Tessman, 2007; Nussey et al., 2014), two employed mixed-methods designs (Wadman, Glazebrook, Beer, et al., 2016; Wadman, Glazebrook, Parkes, et al., 2016), and one study utilised a cross-sectional design (Thomas et al., 2013).

Setting and Location

Two studies focused on only primary school settings (Holtz & Tessman, 2007; Nussey et al., 2014). Four studies focused on secondary school settings only (Fine, 2020; Gilman et al., 2005; Wadman, Glazebrook, Beer, et al., 2016; Wadman, Glazebrook, Parkes, et al., 2016). One study was set across both mainstream primary and secondary schools (Ludlow et al., 2022), and the settings for the last two studies were survey based (Thomas et al., 2013; Grace & Russell, 2005). The location in which studies collected data varied; three studies in the United States of America (USA; Fine, 2020; Gilman et al., 2005; Holtz & Tessman, 2007), four studies in the UK (Ludlow et al., 2022; Nussey et al., 2014; Wadman, Glazebrook, Beer, et al., 2016; Wadman, Glazebrook, Parkes, et al., 2016), one study collected international data from both USA and Australia (ASTL; Grace & Russell, 2005), and one study conducted in Canada (CA; Thomas et al., 2013).

Population

The common populations employed in the included studies were: CYP with TS (or TS+), parents/caregivers of CYP with TS, schoolteachers/staff, and typically developing classmates/students; although samples varied in configuration such as to gain multiple perspectives. Three studies included samples of schoolteachers (Fine, 2020; Ludlow et al., 2022; Wadman, Glazebrook, Parkes, et al., 2016). One study included a sample of only typically developing elementary school students (Holtz & Tessman, 2007). Studies that adopted samples using multiple-perspectives ranged from: one study with a sample consisting of CYP with TS and their parents (Grace & Russell, 2005), two studies with samples comprised of CYP with TS, their parents, and schoolteachers/staff (Thomas et al., 2013; Wadman, Glazebrook, Beer, et al., 2016), and one study with a sample containing CYP with TS, parents, schoolteachers/staff, and typically developing classmates (Nussey et al., 2014).

Out of the nine included studies, only three reported the race and/or ethnicitity of their participants (Holtz & Tessman, 2007; Nussey et al., 2014; Gilman et al., 2005). In Holtz and Tessman (2007), the racial/ethnic backgrounds of their sample of typically developing school children were reported as: 65% Caucasian, 8% African American, 5% Asian, 9% Hispanic, 1 American Indian, and 13% reported "other" as their ethnicity. In Nussey et al. (2014), three of the four participants (CYP with TS) were Caucasian, and one participant described as being "mixed-race." The study by Gilman et al. (2005) only had one participant who's ethnicity was reported as Latino. Additionally, Gilman et al (2005) was the only study out of the nine to report on the socio-economic status of their sample/participant.

Out of the five studies that incorporated CYP with TD in their samples, all focused on participants with TS, no other forms of TD were reported in their samples. Four studies included CYP with TS+ presentations (Thomas et al., 2013; Wadman, Glazebrook, Beer, et

al., 2016; Nussey et al., 2014; Grace & Russell, 2005) with ADHD and OCD being the most frequent co-occuring conditions. The single participant in Gilman et al (2005) was reported to have TS without comorbidity.

Table 5

Summary of Core Characteristics of Included Studies

Author(s) & Year of Publication	Publication Type	Design	Setting and Location	Population	Sample Size	Study Aims/Objectives
Fine, 2020	Doctoral Dissertation	Qualitative Design	Secondary Schools, USA	Secondary School Teachers	n=36	To investigate the level of understanding and preparedness of secondary school educators in supporting students with TS; and identify areas where improvements can be made in teacher preparation programs.
Gilman et al., 2005	Peer-reviewed Article	Experimental Design (Single-case)	Secondary School, USA	CYP with TS	(n=1)	Evaluate the effectiveness of a modified habit reversal intervention for reducing motor tics in an adolescent and to determine if the intervention could be successfully implemented in a school setting with the help of an interpreter.
Grace & Russell, 2005	Peer-reviewed Article	Qualitative Design	Survey, USA, ASTL	CYP with TS and their parents	CYP with TS (n=26) Parents (n=34)	To gain insight into the impact of TS on the school experience by exploring the personal experiences of challenges faced by these children and their families, as well as the strategies they use to cope with these challenges.
Holtz & Tessman, 2007	Peer-reviewed Article	Experimental Design (Randomised Control)	Mainstream Primary school, USA	Neurotypical elementary school students.	(n=179)	To develop and evaluate a video-based intervention to increase children's knowledge and positive attitudes toward a peer with TS.
Ludlow et al., 2022	Peer-reviewed Article	Qualitative Design	Mainstream Primary and Secondary Schools, UK	Primary and secondary school teachers	(n=8)	To (a) explore mainstream primary and secondary school teachers' experience, knowledge, and understanding of teaching children with TS, (b) identify the factors that have contributed to or hindered the success of creating an inclusive environment for children with TS in mainstream schools, (c) identify the training needs of teachers related to TS and to find solutions to improve the inclusion of children with TS in mainstream schools.

Table 5 (Continued)

Summary of Core Characteristics of Included Studies

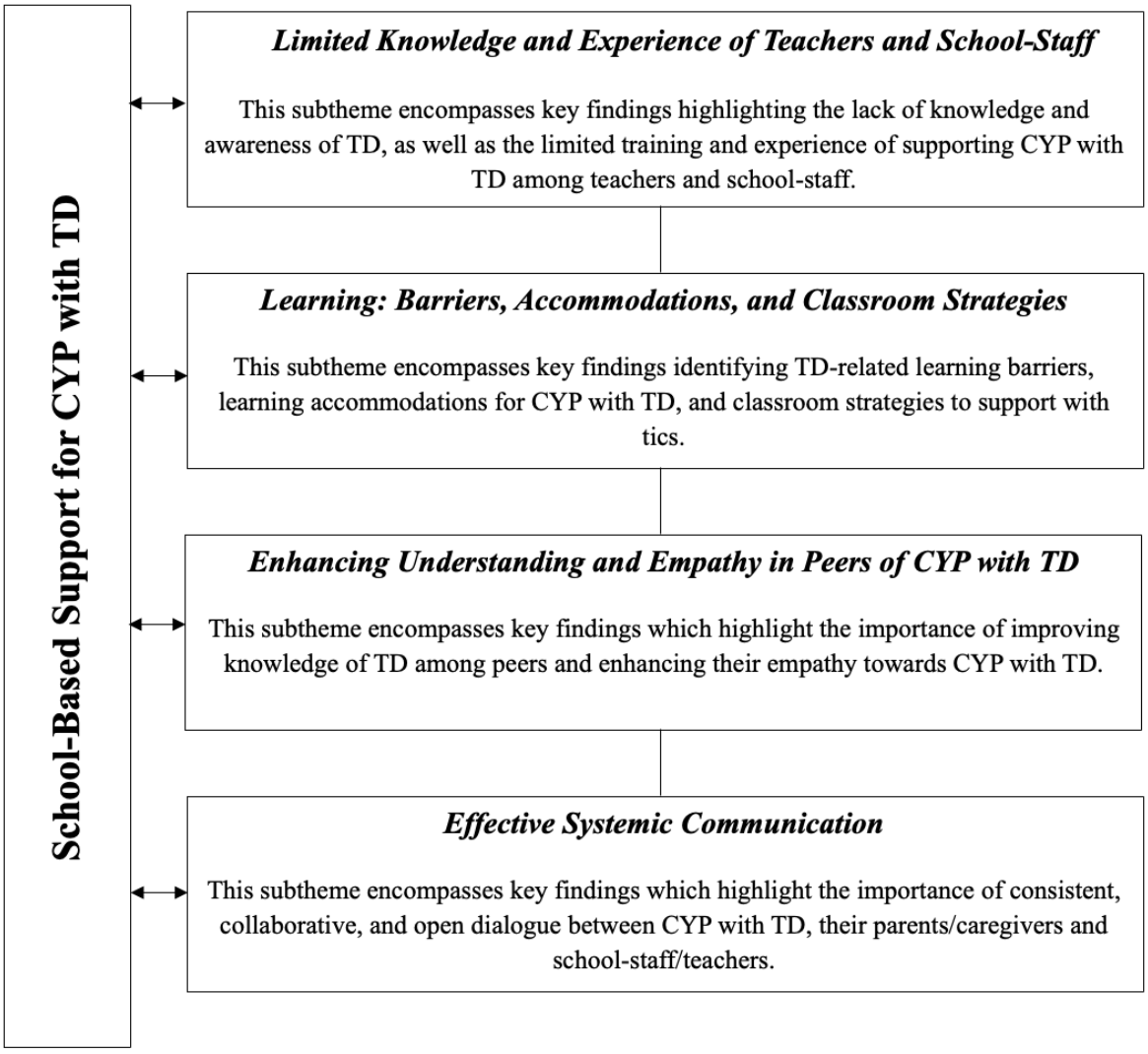
Author(s) & Year of Publication	Publication Type	Design	Setting and Location	Population	Sample Size	Study Aims/Objectives
Nussey et al., 2014	Peer-reviewed Article	Experimental Mixed-Methods Design (Multiple-Case)	Mainstream Primary Schools, UK	CYP with TS, Parents Teachers, and Classmates	CYP with TS (n=4) Parents (n=5) Teachers (n=5) Classmates (n=100)	To (a) explore how children with TS, their parents, teachers and classmates experience the classroom presentation, particularly in relation to the potential impact on the child's peer relationships, (b) to determine whether the child's classmates report a change in knowledge about TS and attitudes towards a child with TS following a presentation.
Thomas et al., 2013	Peer-reviewed Article	Cross-Sectional Survey Design	Survey, CA	CYP with TS+ Parents/caregivers Teachers of children with TS+	CYP with TS+ (n=30) Parents/caregivers (n=30) Teachers (n=20)	To explore the types of educational strategies that these TS+ students (children with TS and other co-morbidities), their parents, and their teachers perceive as most useful for learning, and that children could be taught by significant people in their lives, namely parents and teachers.
Wadman, Glazebrook, Beer, et al., 2016	Peer-reviewed Article	Mixed-Methods Design	Mainstream Secondary Schools, UK	CYP with TS, Parents, and School Staff	CYP with TS (n=35) Parents (n=35) School Staff (n=54)	To (a) explore the difficulties experienced by young people with TS in secondary school, from the perspectives of the young people themselves, their parents, and key members of school staff, (b) inform support strategies by identifying the most common difficulties faced by young people with TS in school, (c) examine the level of agreement between informants and the association with clinical symptom severity.
Wadman, Glazebrook, Parkes, et al., 2016	Peer-reviewed Article	Mixed-Methods Design	Mainstream Secondary Schools, UK	Secondary school staff with responsibilities for special educational needs or disabilities	(n= 63)	To (a) identify support strategies used in schools that could help a student with TS, (b) determine the ease with which a set of recommended strategies could be implemented in schools and to identify any barriers to providing this support.

School-Based Support for CYP with TD: Summary of Key Findings

The key findings from the nine included studies were aggregated into four thematic sub-themes with the overarching theme being school-based support for CYP with TD. Descriptive summaries of the sub-themes are displayed in Figure 2.

Fig. 2

Diagram Summarising Overarching Theme and Sub-themes Used to Report Key Findings



Limited Knowledge and Experience of Teachers and School-Staff

Two studies explicitly explored primary and secondary school teachers' knowledge, experience and understanding of supporting CYP with TD (Fine, 2020; Ludlow et al., 2022). Both studies found that teachers reported receiving little to no information on TD presentations during their professional teaching qualification training; these reports also extended to teachers identified as SENCos and/or teachers who had additional SEN qualifications. These findings highlight that the reported lack and/or absence of training on TD presentations is not specific to the location of where the teachers trained or were working; as one study included teachers in the USA (Fine, 2022), and the other in the UK (Ludlow et al., 2022). Ludlow and colleagues (2022) found that many of the teachers (including SENCos) described TD as the vocalisation of profanities, and reported acquiring much of their knowledge and understanding of TD through the media, which often displays distorted and stigmatising representations of the conditions. Consequently, teachers consistently expressed feeling unequipped and/or underprepared when considering teaching/supporting CYP with TD (Fine, 2020; Ludlow et al., 2022). This lack and/or absence of training was seen to marginalise CYP with TD (Ludlow et al., 2022). Teachers also voiced that due to their lack of knowledge and experience, they were concerned that their response to CYP with TD in school-settings may exacerbate their symptoms. These concerns are made real when considering key findings from studies which included the experiences of CYP with TD and their parents.

In two studies, CYP with TD reported that unhelpful responses and encounters with teachers and school-staff are amongst the most significant difficulties they face (Wadman, Glazebrook, Beer, et al., 2016; Grace & Russell, 2005). In both studies, CYP with TD reported instances of unhelpful responses from teachers, such as being verbally disciplined by teachers in front of classmates or ejected from the classroom; often in response to vocal tics

being considered inappropriate behaviour/attention-seeking. Furthermore, in both studies CYP with TD reported that it generally exacerbated their symptoms when teachers drew attention to their tics. In these instances, Grace and Russel (2005) reported that in almost all the CYP with TD in their study, their sense of tension, embarrassment and ostracism increased as well. A common finding highlighted in the two studies was that the positive attitude of the teacher not only establishes a tolerant classroom environment, but is vital to providing a positive experience for CYP with TD in school-settings (Wadman, Glazebrook, Beer, et al., 2016; Grace & Russell, 2005). Grace and Russel (2005) found that the CYP with TD who were the most pleased with their school experience and academically successful, were those who felt their teachers understood them, their difficulties, feelings, and responded respectfully and sensitively to their needs. Although the findings from studies clearly illustrate their significant lack of knowledge and experience due to limited access to training, teachers commonly recognised this limitation whilst expressing a willingness to learn and engage in finding solutions (Fine, 2020; Ludlow et al., 2022; Wadman, Glazebrook, Beer, et al., 2016). Findings from Wadman, Glazebrook, Parkes, et al. (2016) further reflect this, showing that teachers most frequently recommended staff training when asked about ways of improving support for CYP with TD.

Learning: Barriers, Accommodations, and Classroom Strategies

Findings across five studies identified various barriers to learning CYP with TD face (Wadman, Glazebrook, Beer, et al., 2016; Grace & Russell, 2005), accommodations to support learning, and classroom strategies to support tics (Grace & Russell, 2005; Wadman, Glazebrook, Parkes, et al., 2016; Thomas et al., 2013, Gilman et al., 2005).

Two studies explored and identified frequent barriers to learning reported by CYP with TD. These included difficulties with concentration, handwriting, reading, completing homework and examinations (Wadman, Glazebrook, Beer, et al., 2016; Grace & Russell,

2005). Both studies found that anxiety experienced by CYP with TD significantly exacerbated their tic symptoms and TD-related learning difficulties. Wadman, Glazebrook, Beer, et al. (2016) found that CYP with TD's concentration is not just impacted by the expression of tics (which are intrinsically distractive); but the efforts involved in actively suppressing their tics in classroom settings was reported by CYP with TD to significantly impact their concentration and ability to stay on task. In both studies, CYP with TD reported that motor tics interfered with their handwriting ability. Specifically, Wadman, Glazebrook, Beer, et al. (2016) found that greater severity in motor tics were significantly associated with greater difficulties with handwriting and homework; and the same association was found between phonic (vocal) tics and difficulties with concentration and completing examinations. It was found that when these TD-related learning barriers were experienced as being responded to unhelpfully, or not at all, school avoidance and/or refusal became more prominent in CYP with TD (Wadman, Glazebrook, Beer, et al., 2016; Grace & Russell, 2005).

Findings across all five studies found providing CYP with TD with flexible accommodations and school-based support that is sensitive to their unique TD presentation is essential to facilitating a positive school experience and improving academic achievement (Wadman, Glazebrook, Beer, et al., 2016; Grace & Russell, 2005; Wadman, Glazebrook, Parkes, et al., 2016; Thomas et al., 2013, Gilman et al., 2005). Across three studies, key findings addressed accommodations to support CYP with TD-related learning barriers (Wadman, Glazebrook, Parkes, et al., 2016; Thomas et al., 2013), and school-based strategies to support in managing their tics (Wadman, Glazebrook, Parkes, et al., 2016; Thomas et al., 2013, Gilman et al., 2005). In their study involving secondary SEN school staff in the UK, Wadman, Glazebrook, Parkes, et al. (2016) identified a series of accommodations and school-based strategies SEN staff reported as easily implementable to support learning and school-

based management of tics in CYP with TD. The key learning accommodations they reported included: adjustments for classwork and assignments (e.g. extra time, printed worksheets, use of laptops, reduced homework load, flexible deadlines), and adjustments for examinations (e.g. providing extra time and/or a separate room). These findings are consistent and further supported by those from Thomas et al. (2013) who reported 13 accommodations that were highly endorsed by CYP with TD+. The only variance between the two study's findings regarding types of learning accommodations was the allowance for calculators, spell-checkers, and personalised feedback on work which includes guidance on how to improve (Thomas et al., 2013).

Key findings on strategies to support CYP with managing their tics in school-settings consistently included: teachers not responding to and/or drawing attention to tics, providing an identified safe space for CYP with TD to use when needing to release tics, having teachers/school-staff be educated on TD and provided personalised information regarding each CYP's unique TD presentation and needs (Wadman, Glazebrook, Parkes, et al., 2016; Thomas et al., 2013). Other helpful strategies reported in findings from Wadman, Glazebrook, Parkes, et al. (2016) included: the conscious arrangement of classroom seating for CYP with TD to facilitate both, room for CYP to tic and/or easily exit the classroom if tics become too intense, and allowing CYP with TD to use fidget objects. Findings from the experimental case-design study by Gilman et al. (2005) highlighted the utility and adaptability of implementing behavioural interventions for tics in school-settings. Their findings particularly highlight the benefits inherent in functional analysis (a key component to behavioural interventions for tics). By examining the antecedents and consequences specific to CYP with TD's classroom and/or school-environment, school staff will be better able to understand the variables which might exacerbate, and to some degree, maintain the severity of a student's tics and adjust appropriately. Finally, any accommodations and/or

support strategies which a CYP with TD would perceive as socially unfavourable by their peers will likely be considered as undesirable and lead to further conflict (Thomas et al., 2013).

Enhancing Understanding and Empathy in Peers of CYP with TD

CYP with TD were found to report regularly experiencing victimisation and bullying by peers in school settings. These ranged from teasing and mimicking tics, physical abuse, and resulted in complete social isolation of CYP with TD in most cases (Grace & Russel, 2005; Wadman, Glazebrook, Beer, et al., 2016). Based on their findings, Grace and Russel (2005) reported that when teachers model acceptance and understanding, peers of CYP with TD are likely to emulate this behaviour. Two studies reported findings on psychoeducation interventions aimed at improving CYPs' knowledge and positive attitudes towards peers with TD (Nussey et al., 2014; Holtz & Tessman, 2007). Nussey et al. (2014) found that their classroom-based psychoeducation presentation led to: improved knowledge and attitudes of classmates towards CYP with TD, positively influenced prosocial behaviours in classmates towards CYP with TD, improved self-confidence and facilitated CYP with TD in embracing their condition. Nussey et al. (2014) found their intervention to have a high level of acceptability, with positive qualitative experiences of the presentation being reported by CYP with TD, classmates, teachers and parents. Similar, yet arguably more robust findings were reported in the Randomised Control Trial (RCT) conducted by Holtz and Tessman (2007). Their RCT found that compared to controls, CYP exposed to their peer-focused video-based psychoeducation intervention showed greater improvements in: knowledge, positive attitudes, behavioural intentions and social acceptance towards CYP with TD. Furthermore, it is important to note that through employing an RCT design, the cause-effect relationships found between Holtz and Tessmans' (2007) intervention and outcomes can be considered with greater reliability due to the methodological rigour and robustness inherent in RCT designs.

Effective Systemic Communication

The importance of consistent, collaborative and open dialogue between CYP with TD, their parents/caregivers and school-staff/teachers was highlighted in findings from four studies (Grace & Russell, 2005; Wadman, Glazebrook, Beer, et al., 2016; Thomas et al., 2013; Wadman, Glazebrook, Parkes, et al., 2016). Grace and Russell (2005) found that parents of CYP with TD perceived that a significant investment of time was necessary to volunteer for teachers to negotiate with them. These parents felt that educators would not lend an attentive ear to their concerns purely out of genuine motivation to learn about and support their child; they felt required to demonstrate their willingness to offer their time in return for this benefit. Furthermore, families from Grace and Russells' (2005) study described an ideal framework in which CYP with TD, their parents/caregivers and teachers regularly engaged in discussions in which each individual is provided an opportunity to express their concerns, and then collaboratively develop strategies and plans for support. Wadman, Glazebrook, Beer, et al. (2016) found that staff report significantly fewer TD-related school difficulties compared to CYP with TD and their parents, highlighting how effective systemic communication may facilitate the identification of problem areas and suitable approaches to support. In Thomas et al. (2013), parents of CYP with TD expressed a desire for communication with teachers, endorsing strategies that would enable parents and teachers to communicate, develop, and evaluate the effectiveness of support strategies between school and home; providing synergy among the systems of CYP with TD. SEN teachers were also found to suggest similar approaches, recommending strategies they rated as highly feasible, such as: arranging regular communication between school and home, seeking information about TD from parents, and collaboration with CYP with TD and their parents/caregivers to ensure the sensitivity of support strategies (Wadman, Glazebrook, Parkes, et al., 2016).

Discussion

The present scoping review and narrative synthesis aimed to: (a) map the characteristics of the included empirical studies, and (b) identify what the empirical evidence reveals about strategies for supporting CYP with TD in school settings, and any emerging gaps in knowledge.

Nine studies were included in the present review; three of which used qualitative designs, three adopted varying forms of experimental designs, two employed mixed-methods designs, and one study utilised a cross-sectional design. Only three studies reported the racial and/or ethnic backgrounds of their participants. The settings of the studies mainly included mainstream primary and secondary schools. The locations in which studies collected data varied; three studies originated from the USA, four studies from the UK, one study from CA, and one study collected international data from both USA and ASTL. Populations included in the nine studies shared similarities and also varied in configuration. Three studies included samples of schoolteachers, and one study included a sample of only typically developing elementary school students. Four studies included a range of combined populations to examine multiple-perspectives: one study included a sample of CYP with TS and their parents, two studies included CYP with TS, their parents, and schoolteachers/staff, and one study's sample consisted of CYP with TS, parents, schoolteachers/staff, and typically developing classmates. Importantly, out of the five studies that incorporated populations of CYP with TD, all five focused on participants with TS or TS+, no other forms of TD were reported to be included in their samples. However, this is unsurprising as TS is suggested to be both the most debilitating and widely recognised form of TD (Szejko et al., 2022). Additionally, a core distinction between the three TD diagnostic categories in the DSM-5 (APA, 2013) concerns the duration of tic symptomology, rather than major differences in clinical characterstics. However, the similarities in clinical characteristics does not

necessarily mean that mean that the experiences of CYP with TS and those with other forms of TD are the same. Thus, a potential gap in knowledge is highighted by the lack of inclusion of CYP with forms of TD other than TS in empirical research.

The limited sample of included studies in this review also illuminates the paucity of empirical research in this area, particularly in respect to research employing experimental designs. In this review, only one study employed a methodologically robust experimental design using RCT methodology. It is important to note this scarcity of evidence, as CYP with TD are estimated to account for 3% of mainstream students worldwide (Adams et al., 2023).

The empirical evidence reviewed in this study makes clear that teachers (including SENCos) and school-staff require training to improve their awareness and understanding of TD in CYP. This is also in line with reports of post-graduate SEN programmes varying greatly in regards to topics covered in curriculum (Esposito & Carroll, 2019). CYP with TD will likely require accommodations to help manage TD-related learning difficulties, and findings in this review highlight learning accommodations and classroom strategies that have been identified as useful by not only CYP with TD, but their parents and teachers as well. The present review's findings shed light on the benefits of Psychoeducation groups in enhancing knowledge and empathy in both teachers and peers towards CYP with TD. Finally, key findings from the empirical evidence highlight the importance of synergy among the systems that CYP with TD exist within. Moreover, they place emphasis on collaboration; asserting that together, systems can be a powerful driver which can either positively influence or adversely impact the development and well-being of CYP with TD.

Strengths and Limitations

For the purposes of exploring what empirical evidence reveals about school-based support for CYP with TD, the present review only included studies which reported *primary* data. Thus, the sample of included studies was both limited and heterogenous. However, the

heterogeneity of the studies may also be considered a strength. This paper reviewed quantitative findings as well as including the qualitative perspectives of CYP with TD, parents/caregivers, and teachers; all of whom are critically interdependent, and require symbiosis in order to positively influence the development of CYP with TD (Grace & Russell, 2005; Wadman, Glazebrook, Beer, et al., 2016; Thomas et al., 2013; Wadman, Glazebrook, Parkes, et al., 2016). In accordance with current methodological guidance for scoping reviews (Pollock et al., 2023), no quality assessments were applied to the included studies. As stated in their guidance, "scoping reviews are descriptive in nature; they aim to map the available evidence or identify characteristics or factors" (Pollock et al., 2023, p. 525). The present scoping review's main objective was to provide an overview rather than in depth analysis of empirical findings in the area of school-based support for CYP with TD. However, it is important to note that empirical evidence in the present review was largely descriptive, with only three studies employing varying experimental designs (Gilman et al., 2005; Holtz & Tessman, 2007; Nussey et al., 2014), and only one utilising a randomised control group (Holtz & Tessman, 2007).

Implications and Future Research

The findings from this review offer practical implications which can be used to improve the school-based support of CYP with TD. In light of the limited knowledge, training and experience of TD among teachers (including SENCos), it is essential that training programmes for teachers and post-graduate SEN qualifications review their mandatory curriculum to include training on identifying and supporting TD in CYP. Additionally, any learning accommodations and/or support strategies offered in school should be sensitive to individual needs of CYP with TD, as presentations are fluid and tics fluctuate. Regular dialogue and collaboration between CYP with TD, their parents/caregivers and school is vital to fostering an environment that is inclusive and facilitates positive learning

and social experiences. Psychoeducation presentations on TD should be recommended for both teachers and peers, as these are shown to increase understanding of TD, and prosocial attitudes and behaviours of peers towards CYP with TD. Future research should incorporate populations which are inclusive of other forms TD rather than TS-specific; despite the similarities in clinical characteristics between TD categories, the experiences among individuals may differ between forms of TD.

Conclusion

The present scoping review provides a comprehensive mapping of empirical evidence and digest of their respective key findings related to school-based support for CYP with TD. Moreover, this study's mapping of empirical evidence illuminates the dearth of empirical research in this area, particularly experimental studies. However, this review's findings highlight important implications for teachers, SEN staff and their respective training programmes. This study effectively illustrates the difficulties CYP with TD face in school settings and constructively outlines the school-based accommodations and support strategies, underpinned by empirical evidence, that can be used to improve their school experience, development, and overall well-being.

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Declarations

Conflict of Interest: We have no known conflicts of interest to disclose.

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Appendices

Appendix A Template of Standardised Data Extraction Chart

Data Extraction Form

Scoping Review Questions:

1. What are the characteristics of the empirical studies?
2. What does the empirical evidence reveal about strategies to support CYP with TD in school settings, and what are the gaps in current knowledge?

Data Extraction
Title:
Author(s):
Publication Type:
Year of Publication:
Country:
Study Aims / Objectives:
Design:
Population / Setting:
Sample Size & Characteristics:
Measures:
Method of Analysis:
Findings:
Conclusions:
Limitations & Future Research:

Eligibility Check	
Does the article report *primary data* with the main focus on supporting C&YP with TS/TD in school settings?	
Does the *primary data* involve supporting populations over 18 years old?	
Does the article report primary data on school-settings below Further Education level?	
Is the study written in English?	
Does the title of the publication have Tourette or Tic Disorders in the title?	
Include/exclude (I/E)	

Chapter Two

Empirical Study

"Defining and Characterising a Model of Care for the Assessment, Diagnosis and Management of Tic Disorders in Children and Young People: A Delphi Study"

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Abstract

Background: The prevalence of Tic Disorders (TD) in Children and Young People (CYP) approaches 3%, indicating TD to be more common than previously believed. However, in the United Kingdom, access to National Healthcare Services with a remit for the assessment, diagnosis and management of TD in CYP remains severely limited. The present study aimed to build consensus among healthcare experts to; define and characterise a realistic service model of care with a remit of assessing, diagnosing, and treating TD in CYP, and identify potential obstacles and facilitators to establishing and implementing said service model.

Methods: A panel of experts ($N = 10$) participated in a three-round e-Delphi study. All surveys were completed online. In Round 1, experts provided free-text responses to 7 open-ended questions. Researchers qualitatively processed responses and generated 28 statements which comprised the Round 2 survey. Participants rated statements on relative agreement and/or importance, items that reached consensus were not included in subsequent survey. Controlled feedback of individual and group responses was displayed for participants in Round 3.

Results: Expert consensus was gained on statements pertaining to; service configuration, constraints and obstacles to service development and delivery, age-range and duration of service input, and interventions for service to offer. A ranking hierarchy identifying prioritised professional roles for newly commissioned services was created.

Conclusions: The present study successfully built consensus among healthcare experts to; define and characterise a realistic specialised service model of care with a remit of assessing, diagnosing, and treating TD in CYP, and identified potential barriers and facilitators to establishing and implementing said service model. Findings also highlight the importance of multi-disciplinary team-working and integrated care. Importantly, the present study further emphasises the need for the systematic development of clinical guidelines for TD by the National Institute of Clinical Excellence.

Keywords: Tics, Tic Disorders, Tourette Syndrome, Children, Young People, Delphi Study

Background

Chronic Tic Disorders (TD), such as Tourette syndrome (TS), are non-curable neurodevelopmental conditions characterized by sudden, persistent, purposeless motor movements, or vocalizations known as tics (1). TD typically have an early onset in childhood, peak during early adolescence and fluctuate in both frequency and severity over time. (2) The prevalence of TD in Children and Young People (CYP) under the age of 18 approaches 3% globally, indicating TD to be more common than previously believed; with prevalence rates in the United Kingdom (UK) that come close to matching those of Autism Spectrum Condition in school-aged children (3). However, NHS services with a remit for the assessment, diagnosis and management of TS and TD in CYP remains severely limited.

Empirical evidence has consistently shown physical and behavioural symptoms of TD in CYP to contribute to stigma and social discrimination, poor health outcomes, overall reduction in quality of life (QoL), poor self-esteem and decreased motivation to seek help (4). Adverse outcomes have also been shown to continue into adulthood, with individuals enduring significant rates of exclusion, not just interpersonally but on a more global level, including life domains such as employment, education and healthcare (5). Early diagnosis and basic intervention such as psycho-education have been shown to have a significant, positive and protective influence on self-perception and QoL in CYP that extends into adulthood (6). However, these benefits cannot be gained without access to informed and reliable specialist healthcare services and professionals.

The processes involved in the assessment, diagnosis and management of TD are known to be complex and difficult to navigate in the UK and within the NHS (7). Patients and parents of CYP with TD describe many primary care health professionals being unfamiliar and/or uninformed about tics, leading to delays in obtaining an assessment and reliable diagnosis (8). Individuals in the UK face significant geographical and systemic

barriers to accessing care, with only a handful of specialist services in England, and no specialist services or care pathways in Wales and Scotland. Moreover, there is significant variation in how care is being delivered to CYP with TD across the UK; particularly in the structure of service delivery models (i.e., standalone services vs integrated care pathways) and types of support they provide service-users (9).

Data from a recent international survey of health care services available to patients with TD provided evidence that highlights the concerning state of service provision for this population within the UK (10). Out of all the surveyed countries, the UK revealed the longest wait times at 3-6 months only after a referral to a specialist service had been made. Only eight specialists, (described as clinicians whose practice included more than 60% of patients with tics) were identified in the UK; and patient respondents in the study reported enduring a long and arduous process before receiving a diagnosis. These results made clear that the combination of long wait times for initial assessments and the limited number of specialist clinicians and services highlights the need for multi-disciplinary team (MDT)-working. There are specific benefits to implementing integrated services with MDTs; such as, increased collaboration, pooled expertise which would improve managing TD and common comorbidities, and shorter wait times (10).

Debates have been held in both the UK Parliament and Welsh Senedd during 2022, highlighting the urgent need for specialist services aimed at providing assessment, diagnosis, and care for CYP with TD. The Welsh Government (2022) published a written statement announcing their decision to make an additional £12 million available to support a new national improvement programme for neurodevelopmental conditions (11). They specifically stated that their "aim is to build on these foundations to ensure equity of services and support for people with other neurodevelopmental conditions, such ADHD and Tourette's Syndrome." These recent decisions along with the NHS Long-term Plans for England (12),

Scotland (13), and Wales (14) highlight the present time as a critical juncture in which collaboration and integration are prioritised in respect to healthcare service development and delivery.

Despite repeated requests and significant efforts made by Tourettes Action UK, healthcare professionals and the general-public, there remains an absence of national clinical guidelines; with the National Institute for Clinical Excellence (NICE) denying the latest request for their development this year. When considering the current need for specialist services for CYP with TD in the UK, the lack of national clinical guidelines means there are substantial uncertainties which the present study will address; such as, service configuration, funding arrangements, operational structure, interventions and barriers and facilitators to service implementation.

Study Objectives

Through expert consensus, the present study aimed to: (a) identify elements which define and characterise a realistic NHS service model with a remit of assessing, diagnosing, and treating TD in CYP, and (b) identify the potential barriers and facilitators to establishing and implementing said service model.

Methods

Delphi Method

The Delphi method is a systematic process of gaining consensus from a panel of experts in a particular field who anonymously participate in a series of iterative rounds of survey data collection and controlled feedback (15). The Delphi method was originally developed by the RAND Corporation during the Cold War (16) and has since been established as a reliable research method in gaining expert consensus to answer complex questions (17). The Delphi method has been applied in many domains of healthcare research, such as: healthcare-related interventions, the formation of clinical and best practice

guidelines (18), and development and improvement of healthcare services and models of care (19-21).

Study Design

The present Delphi study consisted of three survey rounds. In line with current best practice guidelines for conducting and reporting Delphi studies (22), specific criteria were defined a priori. The present study defined consensus as being gained when the percentage of agreement among experts reached 70%, which generally exceeds thresholds from other studies employing similar sample sizes and scales (23). Non-consensus was defined as failure to reach consensus after each round. The stopping criteria was operationalised as the completion of three rounds or if all items met consensus. Criteria for dropping items from survey rounds was defined as being satisfied only if: consensus on an item was reached, or if there were more than one item addressing the same statement (e.g. two different statements on what the service age limit should be) and one reaches consensus; that item and its alternatives would then be dropped. Surveys allowed participants to recommend the inclusion and/or adjustment to the wording of statements. A criterion for this was also defined a priori, requiring at least two similar suggestions for the adjustment or adding of items into a subsequent round.

Ethical Considerations

The present study gained ethical approval from the School of Human and Behavioural Sciences Research Ethics Committee at Bangor University in the UK. All participants provided informed consent prior to any data collection.

Expert Panel

Experts ($N = 10$) were defined in the present study by the eligibility criteria which were also set a priori. Experts were defined as eligible if they met either of the following criteria:

a) Professionals who practice in the UK and have been working clinically (with dedicated/protected clinical time allotted to work involving TS/TD populations) for more than five years.
b) Professionals who have been listed as an author in at least 5 peer-reviewed publications in the field of TS or TD.

Experts were recruited through the following sources: Tourettes Action (leading UK charity), TS clinical guidelines, service and policy steering groups organised by the Centre for ADHD and Neurodevelopmental Disorders at University of Nottingham, an advert (Appendix A) posted to and shared on Twitter and LinkedIn, as well as snowball sampling (experts identified by other participants). Once potential participants emailed the researcher, they would be provided with a participant information sheet and informed consent document; which they signed and returned electronically. The 10 experts recruited in the present study were primarily clinical; with 9 meeting inclusion criteria in group a, and one meeting the criteria in group b.

Data Collection

Data was collected from three Delphi survey rounds over a nine-week period between February 2023 and April 2023. The duration for each round was three weeks in total, with the first two weeks allocated to participants and the final week to researchers for analysis and aggregation of responses used in the development of surveys for subsequent round. Surveys were developed and tested by researchers prior to administration, as well as disseminated and completed by participants online using Jisc Online Surveys platform (www.Jisc.ac.uk). Anonymity of participants and their responses were protected through allocating participant numbers, and custom links for each survey round were emailed to each expert through Jisc. To manage attrition, the landing page for each round displayed the closing date (i.e. deadline)

in large, bold, red font. Personalised reminders for completion were also sent through Jisc at two time-points (seven and three days from closing date).

Round 1 Survey

The first survey opened with a participation information page outlining in detail what each Delphi round would entail, as well as the content from the participant information sheet. Delphi methodology used in healthcare research varies, some researchers endorse conducting literature reviews and constructing surveys for the first round accordingly (24). However, others suggest that using open-ended questions in first round Delphi surveys allow participants to express their own views on an issue, facilitating information gathering which extends beyond what is available in the literature (25). Additionally, this approach simultaneously prevents bias from being introduced to the study by researchers imposing their own views on participants (26). Whilst both are widely recognised as acceptable approaches, the present study's first round survey was comprised of seven open-ended, free-text response questions (Table 1). The rationale for utilising open-ended questions in the first round is consistent with the present study's background and aims; as well as being influenced in part by the paucity of existing empirical evidence, which would be needed for data in order to reliably develop a structured questionnaire. Using experts' practical experience to design subsequent surveys through open-ended responses will enrich the current study's findings; in that, they will inherently reflect the current state of affairs (i.e. NHS constraints, policies, etc.) and relative clinical implications.

The Round 1 survey questions (Table 1) were informed by the principles developed by the National Institute for Health Research (NIHR); which is widely used in the process of designing, implementing and evaluating new NHS models of care (27-28). Participants were instructed to answer each question as fully as possible with explanatory detail; whilst also clarifying that they were not expected to "write an exhaustive report."

Table 1

Open-ended Questions Presented in Round 1 Delphi Survey

1. What kind of service structure (e.g., stand-alone, specialised care-pathway) would you recommend as being best-practice for this population? What would be the main arguments to support this recommendation?
2. What are the potential constraints/obstacles for implementing this service structure?
3. What should be the age constraints for this service?
4. What occupations/Roles should be involved in delivering services?
5. What types of interventions should this service offer?
6. What are your views on the duration this service should provide input for?
7. Are there other questions/issues/areas that you feel should be addressed which aren't covered in the items above?

Round 2 Survey

The Round 2 survey contained a total of 28 statements generated following qualitative processing (see Data Analysis for further detail) of the collective responses from experts in Round 1. Themes that arose for these statements were in line with the principles found in the NIHR (2019) framework, such as: Service Configuration, Constraints and Obstacles to Service Development and Delivery, and Operational Structure of Service Model. For items 1-18, participants were asked to rate their level of agreement with a statement using a 5-point Likert scale (1 = Strongly Disagree, 2 = Disagree, 3 = Neither Agree nor Disagree, 4 = Agree, 5 = Strongly Agree). Participants were also provided with an "Other" option, enabling them to respond with a free-text answer if they felt a statement needed adjusting, or simply to provide a response which they felt required further context. Allowing the option for free-text response has been shown to significantly enhance the quality and relevance of survey content in Delphi studies (29). For items 19-28, participants were presented with a list of professional roles identified in Round 1 responses and asked to rate their essentialness to delivering the

respective service using a 3-point Likert scale (1 = Not Required, 2 = Desirable, 3 = Essential).

Round 3 Survey

The Round 3 survey was comprised of 12 items which did not gain consensus in the previous Round. Based on expert feedback from the previous round, the first statement regarding service configuration was adjusted, and experts were asked to choose the structure they would recommend. Round 3 surveys were individually personalised for each participant in order to facilitate controlled feedback, which is considered an essential component in Delphi research methodology (30). For each statement, participants were presented with an image (Appendix B) displaying their previous response (e.g. in Round 2), alongside the average percentage of responses from the group shown in parentheses. This provided experts with the opportunity to compare, consider, and adjust their response if desired; often bolstering the consensus finding process through the convergence of opinions (31).

Data Analysis

Qualitative

Content from the free-text responses in the Round 1 survey were independently read and analysed by the first and second authors separately. For each question, researchers read through each participant's response, highlighted the frequency, order or intensity of occurrence of words, phrases or sentences, and assigned them to emerging categories. Categories which were similar in meaning were combined into a single statement (see Appendix C for worked example). Categories and subsequent statements were validated through the following process: both researchers discussed their findings i.e., the characteristics and overall meaning of each category, establishing agreement that categories accurately reflected participants' responses and that each statement accurately reflected the

categories they subsumed. All statements generated from this process and included in the Round 2 survey required full agreement between researchers.

Quantitative

Percentage of agreement on Likert-scale rated items in Round 2 and Round 3 was calculated through Jisc Survey Software. The 5-point Likert scale data was trichotomised into 3-points; with disagree representing "1-2," neither agree nor disagree remaining, and agree representing "4-5." Numerical values were given to responses (1 = Not important, 2 = Desirable, 3 = Essential) and summed in order to create a ranking hierarchy identifying prioritised professional roles for newly commissioned services.

Results

A flowchart (Figure 1) was created to illustrate each stage and their respective outcomes of the present study's Delphi process. Delphi statements and their consensus outcomes were organised based on their respective themes and reported in the results (see Tables 1-5, and Appendix D). In Round one, ten participants were invited with 100% completing the survey comprised of 7 open-ended questions. In Round 2, ten participants were invited and 100% completed the survey comprised of 28 statements generated from Round 1 free-text responses. In Round 2, consensus was gained on 15 out of the 28 statements (53.57%) while 13 items failed to gain consensus (10 of which related to essentialness of professional roles). One statement concerning duration of service input which failed to reach consensus (see Appendix D; item 3.1) was not included in the subsequent survey since consensus was reached on its alternative. In Round 3, ten participants were invited with 100% completion. The final survey which was comprised of 12 items which did not gain consensus in the previous Delphi round. In Round 3, eight items gained consensus while four (which pertained to professional roles) did not reach consensus.

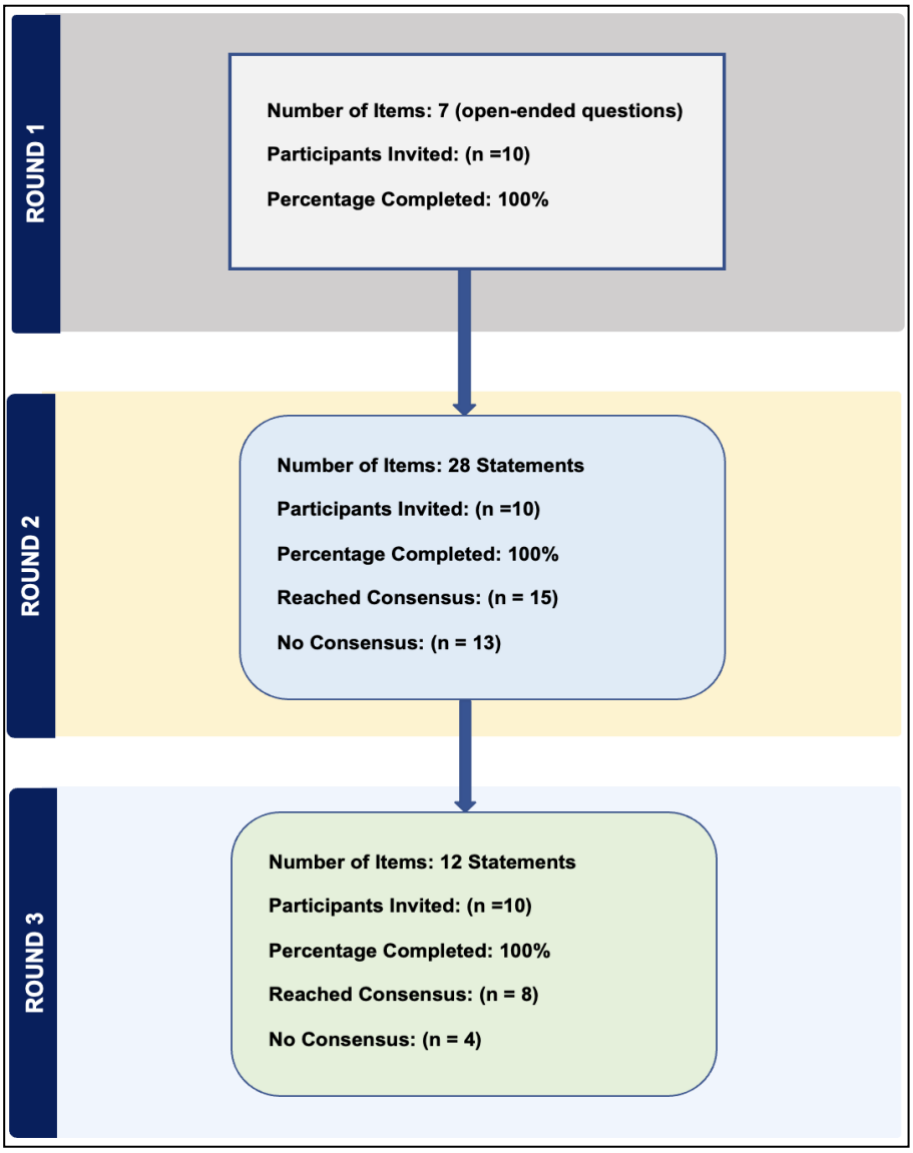

Figure 1. Graphical Flowchart of the Delphi Process

Service Configuration

Consensus on the service configuration statement (Table 2) was achieved in Round 3 with responses being adjusted to forced choice. As this was a key element to the present study, the process of iteration was restricted due to the limited number of Delphi rounds. The first and second author discussed the choice for adjusting response; both agreed that as this was an essential component to the study, and the words/overall message of the statement did not deviate from the original format significantly, using a forced choice response was acceptable as long as it was noted in results.

Table 2

Service Configuration: Question, Statement and Consensus Outcome

When considering the current need in the UK and the development and commissioning of new services, which of the following service models would you recommend?

Delphi Statement	Percentage of Agreement
An integrated specialist care pathway held jointly by community services for Neurodevelopmental and Mental Health conditions (e.g., Paediatrics and CAMHS).	**100 %**

Constraints and Obstacles to Service Development and Delivery

Experts identified and rated the importance of a series of potential constraints and obstacles to developing and implementing this service model. All items (Table 3) in this theme reached consensus in Round 2.

Table 3

Constraints and Obstacles to Service Development and Delivery

How important are these factors when considering the potential constraints and/or obstacles to developing and delivering this service?

Delphi Statement	Percentage of Agreement
Local clinicians (e.g., GPs, Paediatricians, CAMHS practitioners) should have sufficient training using psychometrics to properly assess and diagnose TS & TDs in C&YP.	**80%**
Paediatrics and Mental Health Service Commissioning teams and funding budgets should be integrated.	**80%**
Professional accrediting organisations (e.g., RcPsych, BPS, HCPC) should include training on assessment and diagnosis of TS and TD in their mandated topics.	**100%**
Clinicians at this service should be provided opportunities to engage in clinical research.	**80%**
National Clinical Guidelines should be developed and implemented.	**100%**

Operational Structure of Proposed Service Model

Experts identified and rated their agreement on the importance of various elements concerning the operation of the proposed service model.

Age Range

Consensus on the age range (Table 4) for the proposed service model was achieved in Round 2. This finding was supported by the consensus disagreement on the alternative to the age range experts agreed most with.

Table 4

Operational Structure of Proposed Service Model: Age Range

How important are these factors when considering the operational elements of this service?	
Delphi Statement	**Percentage of Agreement**
This service should have an age limit of 18 years old with structured support for transition into adult services.	**80%**
This service should have an age limit of 16 years old with structured support for transition into adult services.	**70%** (*Consensus Disagreement)*

Duration of Service Input

Consensus on the duration of input (Table 5) from the proposed service model was achieved in Round 2.

Table 5

Operational Structure of Proposed Service Model: Duration of Input

How important are these factors when considering the operational elements of this service?	
Delphi Statement	**Percentage of Agreement**
Within CAMHS age range, this service should have no time limit; service input and discharges should be based on clinical need, with provision to re-engage via GP referral.	**90%**

Interventions

In regard to statements pertaining to interventions for the proposed service to offer (Table 6), all but one statement achieved consensus in Round 2; with the statement relating to TMS reaching consensus in Round 3.

Table 6

Operational Structure of Proposed Service Model: Interventions

How important are these factors when considering the operational elements of this service?

Delphi Statement	Percentage of Agreement
This service should offer Comprehensive Assessment and diagnosis of all presenting neurodevelopmental conditions.	**70%**
This service should offer Psychoeducation for C&YP, families and schools	**90%**
This service should offer Behavioural Therapies such as CBIT, ERP, Habit Reversal	**90%**
This service should offer Psychological Therapies such as CBT and third wave interventions.	**80%**
This service should offer Pharmacotherapy	**80%**
This service should offer Specialist training and consultation clinics for community clinicians and schools (e.g., staff and teachers)	**90%**
This service should offer Non-invasive interventions such as Transcranial Magnetic Stimulation (TMS).	**70%**

Professional Staffing

A ranking hierarchy (see Figure 2) was created to determine which professional roles experts considered most essential to implementing the proposed model of care.

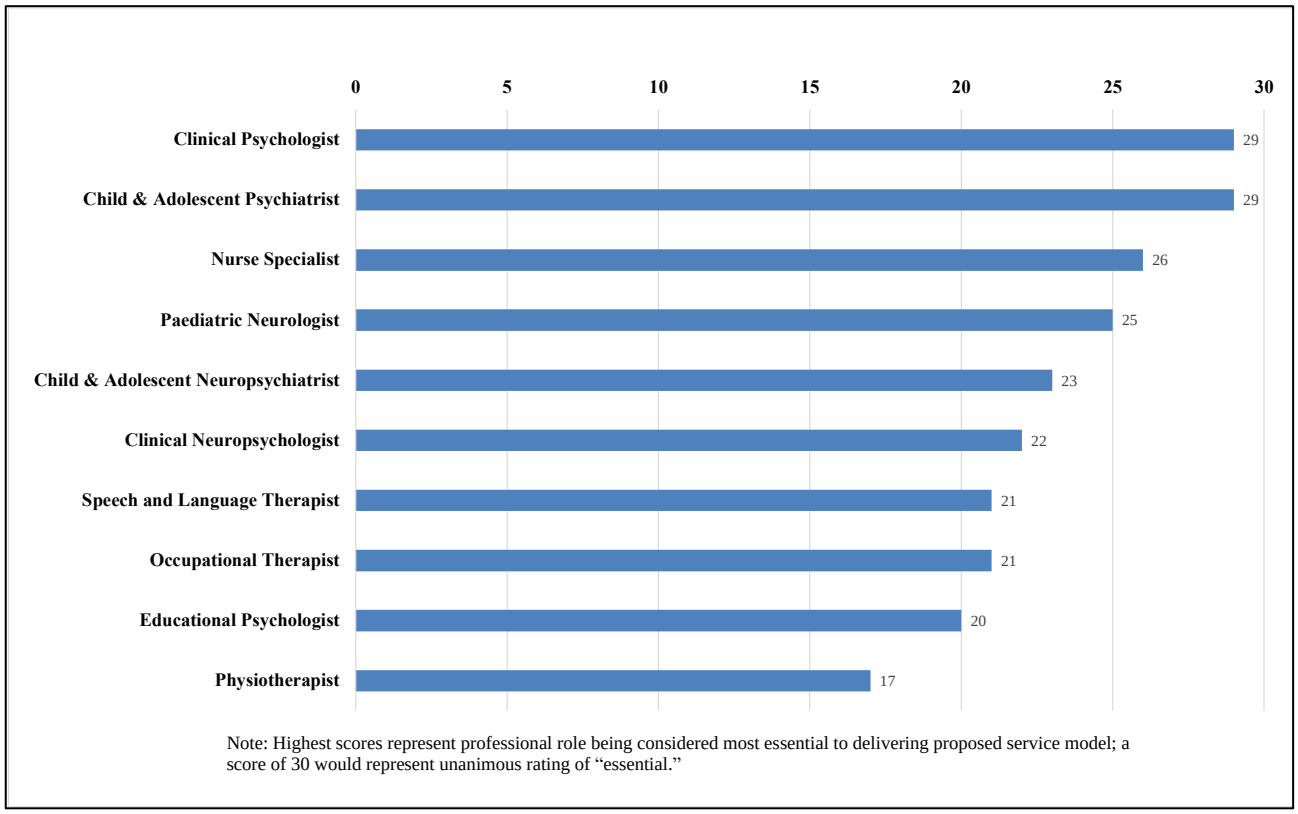

Figure 2 Ranking Hierarchy of Professional Roles.

Summary of Service Model: An Integrated Specialist Care Pathway for CYP with TD

A visual illustration (Figure 3) was as created to summarise the key elements comprising the proposed service model. All elements included in the visual summary achieved consensus among healthcare experts.

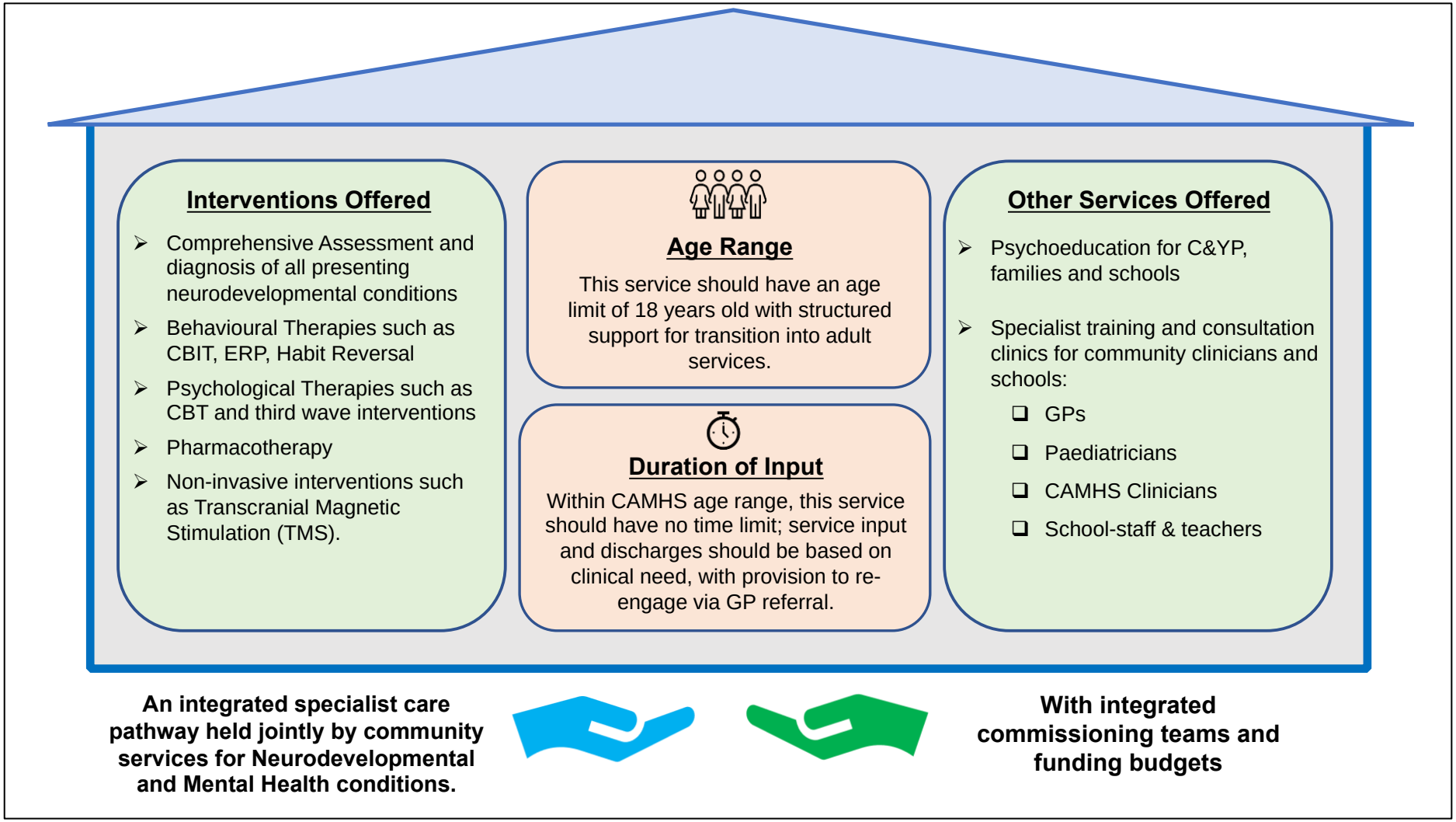

Figure 3 Illustration summarising the key elements comprising the proposed service model.

Discussion

This study utilised a classic e-Delphi design aimed at building consensus among healthcare experts to; define and characterise a realistic NHS service model of care with a remit of assessing, diagnosing, and treating TD in CYP, and identify potential barriers and facilitators to establishing and implementing said service model. To our knowledge, this is the first Delphi study to utilise expert consensus with this intention, and it was successful in identifying and gaining expert consensus on elements which are fundamental to the design and implementation of healthcare models (27). The findings from the present study offers guidance to existing services and clinical pathways, future NHS commissioning teams, policymakers, researchers, and clinicians working with CYP with TD.

This study's findings are in line with existing evidence highlighting the importance of MDTs (evidenced in ranking hierarchy), and the potential benefits of integrated healthcare delivery e.g., avoiding complicated referral processes and increased wait times (10). Furthermore, findings from the present study are consistent with published NHS Long-term plans across England, Wales, and Scotland (12-14); emphasising the importance of integration in service development and delivery. Importantly, experts stressed the cruciality for developing national guidelines (e.g. NICE guidance). The results from this study represents an intentional step towards addressing the absence of NICE guidance for TD. This study's findings are informed by experts who not only understand how our healthcare systems operate nationally, but also reflect their in-depth experience and knowledge of working within the current organisational constraints. This is particularly demonstrated by the experts' consensus on various potential obstacles and relative practical solutions.

Strengths and Limitations

One particular methodological strength to the present study is demonstrated by 100% of experts completing each Delphi Round. Additionally, by the completion of the third round,

expert consensus was reached on all statements pertaining to service configuration, constraints and obstacles to service development and delivery, and the operational structure of the proposed service model (not including professional roles). A potential limitation in this study was its small sample size ($N = 10$). There is no consensus in respect to the optimal size for a Delphi expert panel, although 10-15 has been suggested as sufficient if the backgrounds of experts in a Delphi panel are homogenous like ours was (32). However, it is likely that the completion rates would drop with a larger sample. Additionally, having a smaller and more homogenous pool of experts may have potentially influenced the representativeness of group judgements. However, when considering the variation in percentage of agreement on items across Delphi rounds, this influence does not seem likely.

Although there are limitations to the sample size, there are particular strengths in regard to the sampling approach employed in the present study. First, the robust eligibility criteria the authors set to define healthcare experts provided an added layer of reliability to this study's findings. Second, the present study only recruited experts within the UK, an approach the authors believe fit the purpose of the present study. Recruiting experts within the UK meant that these professionals have an intimate understanding and working knowledge of the current constraints, operations, legislature/policies, and culture associated with the NHS. Therefore, the content developed across the Delphi rounds that comprise the proposed model of care inherently reflected this; and the substantial eligibility criteria these experts met provides additional reliability to the findings.

The present study had a strong rationale for employing open-ended questions in the first round, and it is worth noting that the qualitative process involved in analysing and generating statements was valuable if labour intensive. Considering this, we acknowledge the utility of other approaches used in Delphi studies such as employing focus groups or similar formats in creating statements for the initial Delphi survey (17). Another potential limitation

in the present study related to the ranking of professional roles, despite not all of them achieving consensus. Expert responses to these items may have been affected by the separate Likert-scale used for professional roles. However, in the end the authors felt that creating a ranking hierarchy was equivalent in practice. Finally, although the present study was successful in building expert consensus to define and characterise the proposed model of care, the potential size of the service was not addressed and further details regarding staffing is needed. For example, although professional roles were identified and ranked, information on determining the required number and banding of each professional role based on population need would have been useful guidance to include.

Clinical Implications and Further Research

This study's findings highlight the importance and effectiveness of working in well-designed integrated healthcare systems. Additionally, the results have implications for accrediting organisations responsible for overseeing the curriculum that is used in various professional training programmes (e.g., BPS, HCPC, RcPsych); suggesting training on the assessment, diagnosis and management of TD be included in their mandatory topics. Further research is needed to evaluate the process of implementing the proposed model of care. Some potential outcomes to consider include: staffing/banding based on population need; patient experience; throughflow; level of MDT-working; and effectiveness of protocols and/or treatment approaches.

Although the authors consider the employed sampling approach to be a strength and best fit for the purpose of the present study, it is important to acknowledge the utility of alternative sampling approaches. For example, future research would benefit from the inclusion of international experts. Although there are no existing UK clinical guidelines for TD, there are useful and in-depth clinical guidelines developed internationally (33-34). The present study aimed to define and characterise the content and structure of the proposed

model of care. Consensus was gained on what the service should assess, and which interventions should be offered, but does not account for the clinical procedures and protocols involved in such processes. Thus, including international experts in future research would be beneficial for the alignment of the proposed service model with the current evidence-base.

Conclusion

The present study successfully built consensus among healthcare experts to; define and characterise a specialised integrated NHS service model with a remit of assessing, diagnosing, and treating TD in CYP, and identify potential obstacles and facilitators to establishing and implementing said service model. Importantly, all components related to the proposed service model's configuration, barriers and facilitators to implementation, and operation (i.e., age range, duration of input and interventions offered) achieved expert consensus. The findings from the present study provide an important first step toward addressing the absence of NICE guidelines and the significantly limited access to specialised services for CYP presenting with TD in the UK. Furthermore, the service model characterised in the present study highlights the importance of integrated working, which aligns with NHS Long-term Plans across the UK. Future research should consider evaluating the implementation process of the proposed model of care.

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Appendices

Appendix A Recruitment Advert

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We are currently recruiting healthcare professional experts for a research study which aims to define and characterise what a service model aimed at assessing, diagnosing, and managing Tourette Syndrome (TS) and Chronic Tic Disorders (TD) in children and young people should entail in terms of structure and content.

In the present study, experts are defined as:

- Professionals who have been listed as an author at least 5 peer-reviewed publications in the field of TS or TD, and/or
- Professionals who:
    - Practice in the United Kingdom

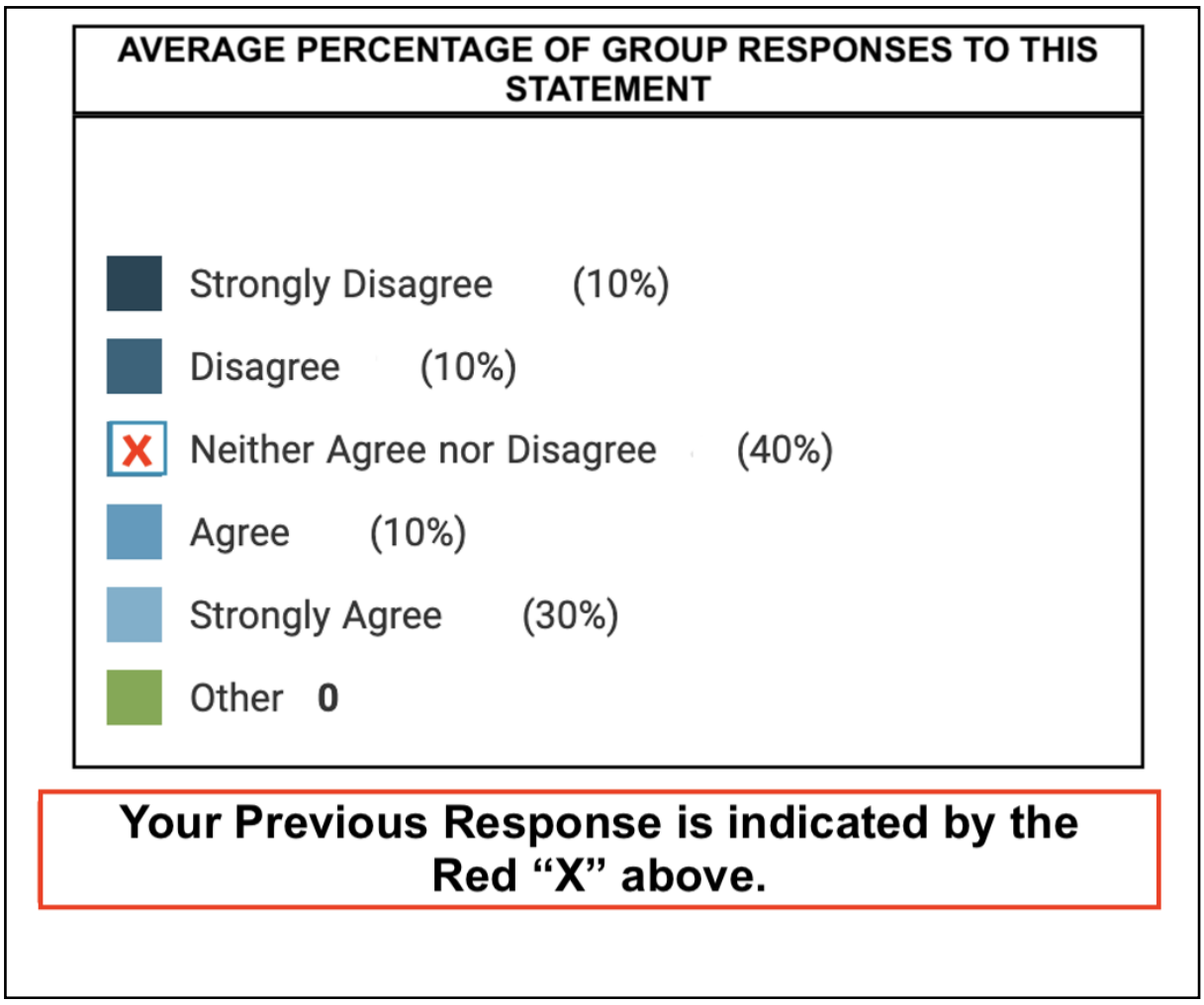
OR

    - Have been working clinically (with dedicated/protected clinical time allotted to work involving TS/TD populations) for more than five years.

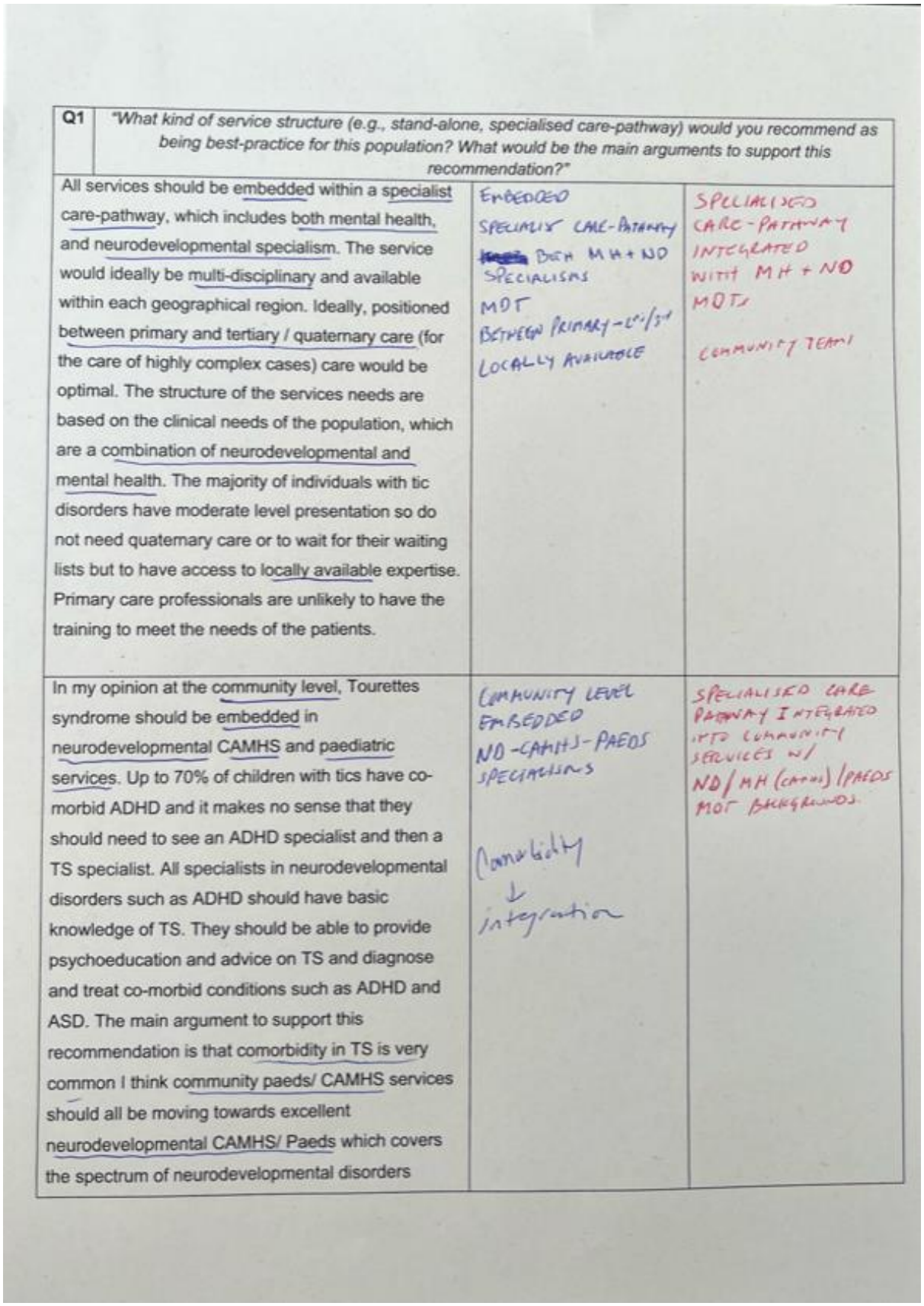
If you meet the criteria above, we would greatly value your contribution! If you are interested in participating and/or just want to hear more about the study, please email Jaxon Kramer (JxK20HKC@Bangor.ac.uk) with answers to the questions below:

1) **Where do you work and what is your current professional role?**
2) **Does your professional experience meet either of the following criteria? If so, please briefly describe how.**
    - **Listed as author on at least 5 peer-reviewed publications in the field of TS or TD.**
    - **Have been working clinically with TS/TD populations for at least five years (with dedicated/protected clinical time allocated to these presentations in work role).**

Appendix B Image Example of Controlled Feedback from Round 3

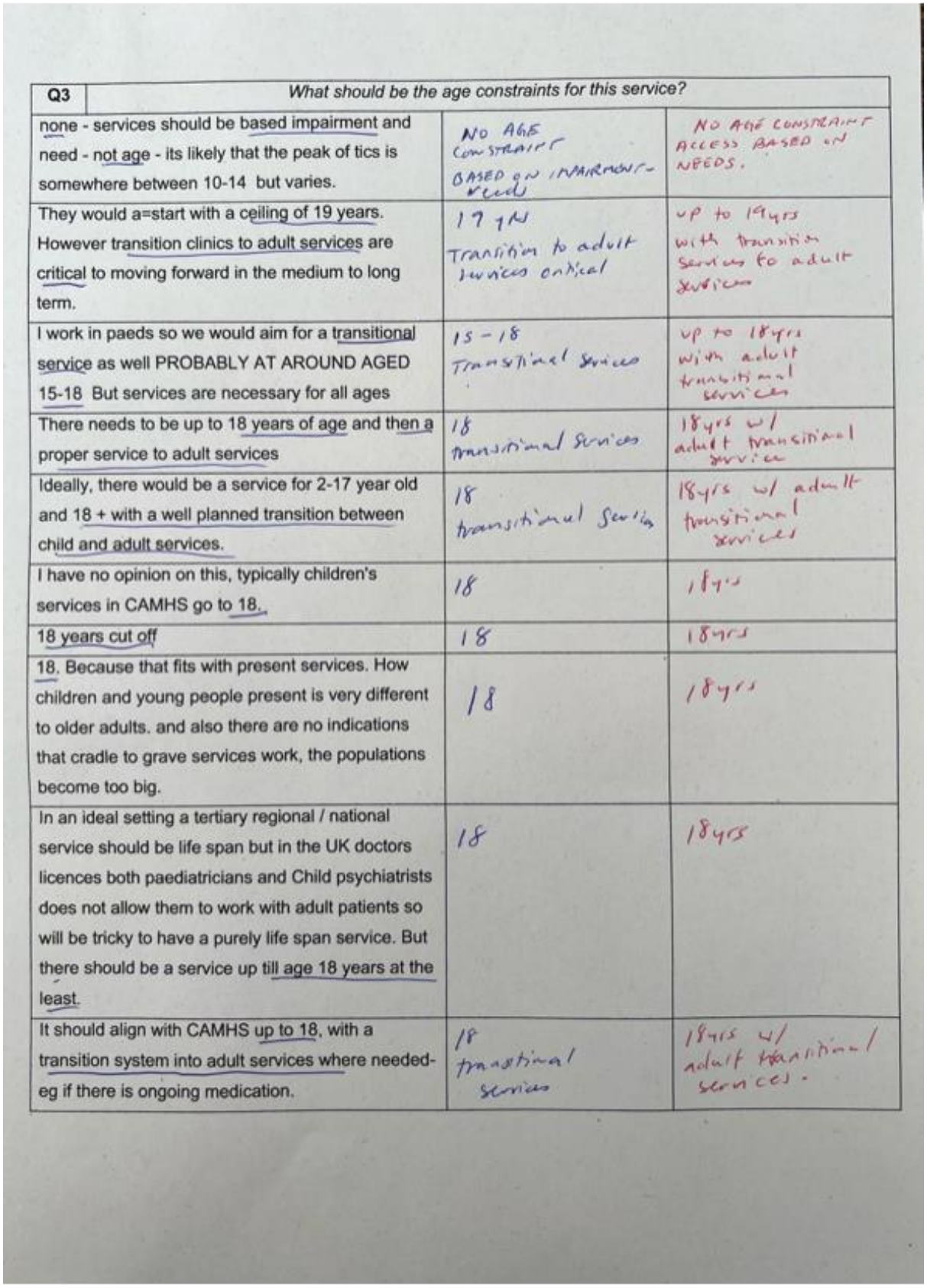


Appendix C Example of Qualitative Processing of Round 1 Responses



Q1	*"What kind of service structure (e.g., stand-alone, specialised care-pathway) would you recommend as being best-practice for this population? What would be the main arguments to support this recommendation?"*	
All services should be embedded within a specialist care-pathway, which includes both mental health, and neurodevelopmental specialism. The service would ideally be multi-disciplinary and available within each geographical region. Ideally, positioned between primary and tertiary / quaternary care (for the care of highly complex cases) care would be optimal. The structure of the services needs are based on the clinical needs of the population, which are a combination of neurodevelopmental and mental health. The majority of individuals with tic disorders have moderate level presentation so do not need quaternary care or to wait for their waiting lists but to have access to locally available expertise. Primary care professionals are unlikely to have the training to meet the needs of the patients.	EMBEDDED SPECIALIST CARE-PATHWAY BOTH MH + ND SPECIALISMS MDT BETWEEN PRIMARY - L²/3ʳᵈ LOCALLY AVAILABLE	SPECIALISED CARE-PATHWAY INTEGRATED WITH MH + ND MDTs COMMUNITY TEAM
In my opinion at the community level, Tourettes syndrome should be embedded in neurodevelopmental CAMHS and paediatric services. Up to 70% of children with tics have co-morbid ADHD and it makes no sense that they should need to see an ADHD specialist and then a TS specialist. All specialists in neurodevelopmental disorders such as ADHD should have basic knowledge of TS. They should be able to provide psychoeducation and advice on TS and diagnose and treat co-morbid conditions such as ADHD and ASD. The main argument to support this recommendation is that comorbidity in TS is very common I think community paeds/ CAMHS services should all be moving towards excellent neurodevelopmental CAMHS/ Paeds which covers the spectrum of neurodevelopmental disorders	COMMUNITY LEVEL EMBEDDED ND-CAMHS-PAEDS SPECIALISMS Comorbidity ↓ integration	SPECIALISED CARE PATHWAY INTEGRATED INTO COMMUNITY SERVICES W/ ND / MH (CAMHS) / PAEDS MDT BACKGROUNDS

Appendix C.1 Further Example of Qualitative Processing of Round 1 Responses



Q3	*What should be the age constraints for this service?*	
none - services should be based impairment and need - not age - its likely that the peak of tics is somewhere between 10-14 but varies.	NO AGE CONSTRAINT BASED ON IMPAIRMENT - NEED	NO AGE CONSTRAINT ACCESS BASED ON NEEDS.
They would a=start with a ceiling of 19 years. However transition clinics to adult services are critical to moving forward in the medium to long term.	19 yrs Transition to adult services critical	up to 19yrs with transition services to adult services
I work in paeds so we would aim for a transitional service as well PROBABLY AT AROUND AGED 15-18 But services are necessary for all ages	15 - 18 Transitional Services	up to 18yrs with adult transitional services
There needs to be up to 18 years of age and then a proper service to adult services	18 transitional Services	18yrs w/ adult transitional service
Ideally, there would be a service for 2-17 year old and 18 + with a well planned transition between child and adult services.	18 transitional Servi	18yrs w/ adult transitional services
I have no opinion on this, typically children's services in CAMHS go to 18.	18	18yrs
18 years cut off	18	18yrs
18. Because that fits with present services. How children and young people present is very different to older adults. and also there are no indications that cradle to grave services work, the populations become too big.	18	18yrs
In an ideal setting a tertiary regional / national service should be life span but in the UK doctors licences both paediatricians and Child psychiatrists does not allow them to work with adult patients so will be tricky to have a purely life span service. But there should be a service up till age 18 years at the least.	18	18yrs
It should align with CAMHS up to 18, with a transition system into adult services where needed- eg if there is ongoing medication.	18 transitional services	18yrs w/ adult transitional services.

Appendix D Tables Listing Each Item and Consensus Outcome for Rounds 2 and 3

ROUND 2: Delphi Survey Items Reaching Consensus	Percentage of Agreement
Q2.1 - Local clinicians (e.g., GPs, Paediatricians, CAMHS practitioners) should have sufficient training using psychometrics to properly assess and diagnose TS & TDs in C&YP.	**80%**
Q2.2 - Paediatrics and Mental Health Service Commissioning teams and funding budgets should be integrated.	**80%**
Q2.3 - Professional accrediting organisations (e.g., RcPsych, BPS, HCPC) should include training on assessment and diagnosis of TS and TD in their mandated topics.	**100%**
Q2.4 - Clinicians at this service should be provided opportunities to engage in clinical research.	**80%**
Q2.5 - National Clinical Guidelines should be developed and implemented.	**100%**
Q.3.1 - This service should have an age limit of 16 years old with structured support for transition into adult services.	**Consensus Disagreement** **70%**
Q3.2 - This service should have an age limit of 18 years old with structured support for transition into adult services.	**80%**
Q4.1 - Within CAMHS age range, this service should have no time limit; service input and discharges should be based on clinical need, with provision to re-engage via GP referral.	**90%**
Q5.1 - This service should offer Comprehensive Assessment and diagnosis of all presenting neurodevelopmental conditions.	**70%**
Q5.2 - This service should offer Psychoeducation for C&YP, families and schools	**90%**
Q5.3 - This service should offer Behavioural Therapies such as CBIT, ERP, Habit Reversal	**90%**
Q5.4 - This service should offer Psychological Therapies such as CBT and third wave interventions.	**80%**
Q5.5 - This service should offer Pharmacotherapy	**80%**
Q5.6 - This service should offer Specialist training and consultation clinics for community clinicians and schools (e.g., staff and teachers)	**90%**
Q6.4 – Clinical Psychologist	**90%**

Appendix D.1 Tables Listing Each Item and Consensus Outcome for Rounds 2 and 3

ROUND 2: Delphi Survey Items That Did *NOT* Reach Consensus	Percentage of Agreement
Q1.1 - This service is a stand-alone design that operates on a tiered or stepped-care system.	**Disagree: 20% Neutral: 40% Agree: 30% Other: 10%**
Q1.2 - This service is an integrated specialist care pathway held jointly by community services for Neurodevelopmental and Mental Health conditions (e.g., Paediatrics and CAMHS).	**Disagree: 10% Neutral: 20% Agree: 60% Other: 10%**
Q4.1a – This service provides input up to 1 year from assessment to completion of behavioural intervention; and if applicable, medication managed/reviewed locally through shared-care plans.	**Disagree: 50% Neutral: 10% Agree: 40%**
Q5.7 - This service should offer Non-invasive interventions such as Transcranial magnetic stimulation (TMS)	**Disagree: 20% Neutral: 40% Agree: 40%**
Q6.1 - Paediatric Neurologist	**Essential: 40% Desirable: 50% Not Required: 10%**
Q6.2 - Child & Adolescent Psychiatrist	**Essential: 60% Desirable: 40% Not Required: 0**
Q6.3 - Child & Adolescent Neuropsychiatrist	**Essential: 40% Desirable: 60% Not Required: 0**
Q6.5 - Clinical Neuropsychologist	**Essential: 40% Desirable: 60% Not Required: 0**
Q6.6 - Educational Psychologist	**Essential: 20% Desirable: 60% Not Required: 20%**
Q6.7 - Nurse Specialist	**Essential: 60% Desirable: 40% Not Required: 0**
Q6.8 - Speech and Language Therapist	**Essential: 20% Desirable: 60% Not Required: 20%**
Q6.9 - Occupational Therapist	**Essential: 30% Desirable: 60% Not Required: 10%**
Q6.10 - Physiotherapist	**Essential: 30% Desirable: 20% Not Required: 50%**

Appendix D.2 Tables Listing Each Item and Consensus Outcome for Rounds 2 and 3

ROUND 3: Delphi Survey Items Reaching Consensus	Percentage of Agreement
Q1. *When considering the current need in the UK and the development and commissioning of new services, which of the following service models would you recommend?* ↳ An integrated specialist care pathway held jointly by community services for Neurodevelopmental and Mental Health conditions (e.g., Paediatrics and CAMHS).	**100%**
Q2. This service should offer Non-invasive interventions such as Transcranial Magnetic Stimulation (TMS).	**70%**
Q4 - Child & Adolescent Psychiatrist	**Essential: 90%** **Desirable: 10%** **Not Required: 0**
Q5 - Child & Adolescent Neuropsychiatrist	**Essential: 30%** **Desirable: 70%** **Not Required: 0**
Q6 - Clinical Neuropsychologist	**Essential: 20%** **Desirable: 80%** **Not Required: 0**
Q7 - Educational Psychologist	**Essential: 0** **Desirable: 100%** **Not Required: 0**
Q9 - Speech and Language Therapist	**Essential: 10%** **Desirable: 90%** **Not Required: 0**
Q10 - Occupational Therapist	**Essential: 10%** **Desirable: 90%** **Not Required: 0**
ROUND 3: Delphi Survey Items That Did *NOT* Reach Consensus	**Percentage of Agreement**
Q3 - Paediatric Neurologist	**Essential: 50%** **Desirable: 50%** **Not Required: 0**
Q8 - Nurse Specialist	**Essential: 60%** **Desirable: 40%** **Not Required: 0**
Q11 - Physiotherapist	**Essential: 60%** **Desirable: 40%** **Not Required: 0**

Appendix E Confirmation of Ethical Approval

E

ethics@bangor.ac.uk <ethics@bangor.ac.uk>

To: Jaxon Kramer

Dear Jaxon,

Your research proposal number 2022-17246
has been reviewed by the [Pre-Aug 2021] School of Psychology Ethics and Research Committee
and the committee are now able to confirm ethical and governance approval for the above research on the basis described in the application form, protocol and supporting documentation. This approval lasts for a maximum of three years from this date.

Ethical approval is granted for the study as it was explicitly described in the application

If you wish to make any non-trivial modifications to the research project, please submit an amendment form to the committee, and copies of any of the original documents reviewed which have been altered as a result of the amendment. Please also inform the committee immediately if participants experience any unanticipated harm as a result of taking part in your research, or if any adverse reactions are reported in subsequent literature using the same technique elsewhere.

Appendix F Consent Form

PRIFYSGOL BANGOR UNIVERSITY

Participant Consent Form

Study Title: "Defining and Characterising a Model of Care for the Assessment, Diagnosis and Management of Tic Disorders in Children and Young People: A Delphi Study"

Researchers Name: Jaxon Kramer

Please initial each box to show that you agree with each statement:

I have read and understood the information sheet provided to me by the named researcher.	☐
I have had time to consider the information I have had the opportunity to ask any questions, which have been answered satisfactorily.	☐
I understand that taking part in this study is voluntary and I have the right to withdraw from the study, without giving a reason.	☐
I understand the data I give whilst taking part in this study is confidential. Although if the researcher becomes concerned about mine, or another person's, well-being they may have to inform my support worker or another agency.	☐
I agree to take part in this study	☐

Participant Name _____

Participant Signature _____ Date _____

Researcher Signature _____ Date _____

Appendix G Debrief Sheet

PRIFYSGOL BANGOR UNIVERSITY

DEBRIEF FORM

Thank you for taking the time to take part in this study and share your expertise.

What were the aims of this study?

The aim of this study was to develop an expert consensus using the Delphi Process on what the content and structure of an NHS Service Model aimed at assessing, diagnosing, and treating Tic Disorders in Children and young people (C&YP) should entail. In order to achieve this understanding you were asked to share your expertise through providing opinions and answers to a series questions and statements. This information will be analysed and form part of a research report. It is the hope that the study will provide knowledge that can improve the development of policy, clinical guidelines and service provision for C&YP with Tic Disorders in the UK.

What do I do if I have any further questions?

If you have any further questions the researcher will be happy to answer them. You can contact the researcher directly. The contact details are:

- Jaxon Kramer - JxK20Hkc@Bangor.ac.uk

If you would like to find out about the results of the study once it is finished, please ask the researcher and they can go through them with you.

What if I am concerned or have a complaint about any aspect of the study?

If you have any concerns or questions, you can contact Jaxon whose details are above. If you remain unhappy about any aspect of the study, or would like to make a complaint, please contact huw.roberts@bangor.ac.uk, manager of the college of Human Sciences at Bangor University.

Thank you again for taking the time to participate in this research!

Appendix H Participation Information Sheet

PRIFYSGOL BANGOR UNIVERSITY

INFORMATION SHEET

"Defining and Characterising a Model of Care for the Assessment, Diagnosis and Management of Tic Disorders in Children and Young People: A Delphi Study"

I would like to invite you to take part in a research study exploring what a service model aimed at assessing, diagnosing and managing Tourette Syndrome and Chronic Tic Disorders in children and young people should entail in terms of structure and content. The information on this sheet provides details about the study, please read it carefully before deciding to take part.

If you have any questions about the information here, you can contact the researcher whose details are at the bottom of this sheet. They will be happy to answer any questions.

Who is carrying out the research?

Jaxon Kramer is a Trainee Clinical Psychologist on the North Wales Clinical Psychology Programme at Bangor University. This study is being undertaken as part of his training and is supervised by Dr Mike Jackson. The project has been approved by the Psychology Research Ethics Committee.

What is the purpose of this study?

The aim of this study is to develop an expert consensus using the Delphi Process on what the content and structure of an NHS Service Model aimed at assessing, diagnosing, and treating Tic Disorders in Children and young people (C&YP) should entail. I hope to use this understanding to provide translatable research which can be used to inform the development of policy, clinical guidance and service planning in the future.

What will happen if I take part?

If you choose to take part, you will be asked to provide responses to a series of questions (split into rounds) sent to you in email format. The duration of each round will be three weeks, with the expectation of no more than four rounds overall.

Chapter Three

Contributions to Theory and Clinical Practice

Followed by Reflective Commentary

Contributions to Theory and Clinical Practice

Collectively, this thesis aimed to increase our understanding and improve the development of systemic approaches to supporting CYP with TD in school and healthcare settings. In this final chapter, the findings from both the scoping review and Delphi study will be collectively discussed in the context of their implications for theory development, clinical practice and future research. The chapter will conclude with a reflective commentary from the first author on the experience of carrying out this large-scale research project.

Implications for Theory Development, Clinical Practice and Future Research

To date, TD are often viewed through a medical model lens, which sees TD symptomology (i.e. tics) as a problem of deficiency which needs to be ameliorated in order for the individual to function normally within society (Malli & Forrester-Jones, 2022). However, it is often the environments surrounding CYP with TD and not TD themselves that are shown to adversely impact CYP with TD's development and overall QoL (Evans et al., 2016). TD have been associated with poor self-esteem and negative self-concept, which are often socially-constructed through the internalisation of the heightened stigmatisation, victimisation, and bullying CYP with TD frequently face (Lee et al., 2019; Kim & Tak, 2020). Adopting the viewpoint of the medical model towards TD in CYP is akin to trying to fit a square peg into a round hole; although you *can* accomplish such a task, it cannot be done without denying the square peg's nature and then compelling it to conform. Thus, the medical model viewpoint fails to account for the distal influences beyond the control of CYP with TD that significantly effect the systems they are embedded in by contributing to stigmatising sterotypes.

TD are often misrepresented in mainstream media and even weaponised by public figures who are supposed to represent us. A recent Channel 4 documentary titled "Britain's

Tourette's Mystery" (2022) contained a significant amount of harmful, inaccurate and misleading information; with the presenter (television star Scarlett Moffatt) going so far as to suggest TS to be an infectious condition, and tics to be contagious (Tourettes Action, 2022). In 2012, the then UK Prime Minister David Cameron described facing the shadow chancellor Ed Balls in parliament to be like "having someone with Tourette's sitting opposite you" (Reuters, 2012). Research has shown that these types of misrepresentations and harmful portrayals of TD reinforces stereotypes and exacerbates stigmatising attitudes and behaviours (Cox et al., 2019). Therefore, the findings from this research project emphasise the importance of viewing CYP with TD through a *systemic* lens.

The Delphi and scoping review methodologies share an underlying commonality; they were employed because of the general neglect of this topic area in terms of empirical research, clinical guidelines, and accessibility to specialised services. The two studies provide findings which pertain to two key interactive and influential systems which surround CYP with TD: school and healthcare. When incorporated with the Bioecological Model of Human Development (BMHD; Bronfenbrenner & Morris, 2006), the findings from both studies expands our understanding of how and why school and healthcare systems can improve the way CYP with TD are systemically supported in the UK. The findings from the two research papers will be organised using the BMHD model to demonstrate: (1) the value of viewing TD in CYP through a systemic lens, and (2) their implications for theory development, clinical/educational practice, and future research. Although the BMHD can be applied to all children, this paper will specifically discuss the model in the context of CYP with TD.

The BMHD places CYP with TD at its centre, recognising that their development is influenced by reciprocal relationships with the complex network of interconnected systems of which the child exists within. The BMHD identified five systems that surround the child, below is a description of each system and how the implications of this project's findings on

clinical practice and future research theory align with them. Thus, simultaneously demonstrating their collective contribution to the development of theory.

Microsystem

This component refers to the immediate environments and events in which CYP with TD directly interact with, such as family, school, and peer groups. These systems have been evidenced to significantly influence the development and QoL of CYP with TD (Goldsmith, 2016). Findings from our scoping review provide important insights into the interplay within these systems and offers important practical recommendations for improving support for CYP with TD. Various barriers to learning CYP with TD face were identified in our review, such as: difficulties with concentration, handwriting, reading, completing homework and examinations. Additionally, we found a significant lack and/or complete absence of TD-related knowledge and training reported by teachers and SENCos in the UK (Ludlow et al.,2022). This correlates with other findings from paper one in which CYP with TD reported that unhelpful responses and encounters with teachers and school-staff are amongst the most significant difficulties they face (Wadman, Glazebrook, Beer, et al., 2016; Grace & Russell, 2005). CYP with TD were also found to report regular experiences of victimisation and bullying by school peers; ranging from teasing and mimicking tics, physical abuse, and resulted in complete social isolation in most cases. Importantly, findings from our review highlight the benefits of school-based psychoeducation interventions, such as: increasing CYP with TD's self-esteem and ownership of their condition, increasing knowledge of TD in both teachers and peers, and positively influencing peer-attitudes and prosocial behaviours towards CYP with TD.

Implications for Practice and Future Research: The Microsystem

Concerning the limited knowledge, training, and experience of TD among teachers (including SENCos), it is essential that training programmes for teachers and post-graduate

SEN programmes review their mandatory curriculum to include training on identifying and supporting TD in CYP. Teachers and school-staff of CYP with TD need to model acceptance and understanding, as evidence suggests peers of CYP with TD are likely to emulate this behaviour. CYP with TD will likely require accommodations to support with TD-related learning barriers. Any learning accommodations and/or support strategies offered in school should be sensitive to individual needs of CYP with TD, as presentations are fluid and tics fluctuate. Psychoeducation presentations on TD should be recommended for both teachers and peers, as these are shown to improve knowledge of TD, prosocial attitudes, and behaviours of peers towards CYP with TD. Our scoping review identified a paucity of empirical studies in this area, thus highlighting the need for further research. Only one experimental study identified in our review was an RCT design, therefore future research on the effectiveness of school-based interventions should employ RCT methodology as this will strengthen the reliability of findings.

Mesosystem

The mesosystem involves the interactions and connections between CYP with TD's microsystems. It recognises that the experiences and dynamics within one microsystem can impact another, emphasising the importance of coordination and communication across settings. Findings from our Delphi study and scoping review both highlight the importance of integration and collaboration. Similar to implications for training found in our scoping review, the consensus among healthcare experts in the Delphi study highlighted the need for professional training programmes (e.g., BPS, HCPC, RcPsych) to include training on assessment, diagnosis and management of TD in their mandatory curriculum. Furthermore, experts were unanimous in their recommendation that the structure of the proposed service model be integrated, endorsing MDT-working. A key theme within the findings from our scoping review highlighted the importance of effective systemic communication. Evidence

showed that communication and collaboration between parents of CYP with TD and their schools was often challenging. The proposed service model from our Delphi study has the potential to positively influence these microsystems in important ways.

Implications for Practice and Future Research: The Mesosystem

The proposed service model developed from expert consensus can positively influence other microsystems that surround CYP with TD through collaboration. First, professionals from the specialised service are in a prime position to not only inform the development and implementation of school-based interventions and support strategies for CYP with TD; but can effectively evaluate outcomes and work with microsystems to tailor school-based support they are providing. Findings from our Delphi study demonstrate how the healthcare system can improve the synergy within the mesosystem. For example, experts were in consensus that the proposed service should offer interventions such as: psychoeducation for CYP with TD, families and schools; and specialist training and consultation clinics for staff and teachers. Further research is needed to evaluate the process of implementing the proposed model of care. Additionally, outcomes such as the degree of collaborative working among mesosystems would be valuable.

Exosystem

The exosystem involves the external environments that indirectly influence an individual's development. These include community structures, organisations, and social policies. The exosystem recognises the indirect yet influential impact of these external factors on an individual's experiences. The findings from the Delphi study highlight the need for national clinical guidelines for TD. The prevalence of TD in school-aged children is estimated to be approximately 1% in the UK (Hall et al., 2022). Despite the significant efforts made by Tourettes Action UK, healthcare professionals and the general-public, the absence of

national clinical guidelines remains; with NICE denying the latest request for their development this year.

Implications for Practice and Future Research: The Exosystem

The development of NICE guidance for TD can benefit CYP with TD's entire *ecosystem*. For example, the benefits of implementing NICE guidelines can range from: providing clinicians with easily accessible means to best practice, improving planning and commissioning of NHS services, increasing opportunities for allocated resources, to improving the economic and social well-being of communities (NICE, 2023). Currently, access to specialist services for CYP with TD is severely limited in the UK, with minimal funding for the commissioning of new services (Bhikram et al., 2021). NICE guidelines are also shown to inform policies related to various health conditions. Therefore, the creation of NICE guidance for TD is essential and can have a positive and wide-ranging systemic influence that benefits CYP with TD.

Macrosystem

The macrosystem encompasses the broader cultural and societal influences that shape development. It includes social norms, cultural values, economic systems, and political ideologies. The macrosystem recognises the overarching context within which the other components of the model operate. The papers from this research project focused on two specific systems, however what isn't addressed is how we can intervene on a societal level. As mentioned earlier in this chapter, CYP with TD are often harmed by the systemic impact of stigmatising attitudes and stereotypes facilitated and maintained through distal influences (Cox et al., 2019).

Implications for Practice and Future Research: The Macrosystem

Again, the importance of creating NICE guidelines for TD is highlighted as they can have a systemic influence on the macrosystem, such as: increasing access to funding,

improving the visibility of TD and the positive impact this can have on legislature. As a product of improved legislation, TD populations become more protected, better understood, and thus knowledge and acceptance of TD will progressively be reflected more in culture and society. Future research would benefit from exploring the impact of public campaigns on public knowledge and attitudes towards TD. For example, Tourettes Action UK holds an annual public campaign "Tourette's Awareness Month" between 15th of May to 15th of June. The aim is to dispel stigmatising myths and stereotypes that surround the condition.

Chronosystem

The chronosystem acknowledges the dimension of time and temporal changes. It highlights how development is influenced by historical events, transitions, and individual life experiences over time. The chronosystem underscores the dynamic nature of development. TD are shown to have a fluctuating nature, and tic symptomology is shown to wax and wane over time (Szejko et al., 2022). These peaks and troughs in tic severity may disrupt the homeostasis and synergy among the systems surrounding CYP with TD.

Implications for Practice and Future Research: The Chronosystem

The temporal fluctuation of tic symptomology commonly observed in TD populations suggests that support provided by healthcare and school systems for CYP with TD will likely require recurrent adaptations to ensure that the support strategies being offered are appropriate to their TD-related needs. Additionally, it is important to note that the implications described across the systems above, underpinned by findings from this research project, have the potential to systemically influence CYP with TD's self-esteem, self-concept and ownership of their condition, which can protect against various adversities over time (Eapen et al., 2016).

Reflective Commentary

Carrying out this research project has been both personally rewarding and challenging for me. My story was echoed in the qualitative accounts reported in many of the studies I reviewed. When conducting the scoping review, memories arose that I hadn't thought of for decades, and others that have stuck with me for my entire life. When I was six years old, my mother told me that one morning she was walking me into nursery school when I paused, looked up at her and said "Mamma, my body does things my mind doesn't want it to do." Within a short span of time, I was seen by a specialist and diagnosed with Tourette Syndrome. I find myself writing this commentary with tears in my eyes, because 30 years later I still remember what it was like to grow up feeling as if I was a stranger in my own body. I was a child overwhelmed by confusion, fear, and anger. I have clear memories of watching the other children smiling and laughing as they played during recess and wondering why they would all pretend to be so happy; because my own experience meant I fundamentally did not understand how that was a possibility. Over the course of 10 years following my diagnosis: I would try countless medications, some would make me sleep all day, some would make me cry for no apparent reason, others would make me irrationally angry, lose weight, and one would make me gain over two stone in a month. My tics would cause my kneecaps to start chipping, I would find myself isolated, cut-off from the world and afraid to leave the house due to my coprolalia which would cause me to scream racial slurs. I would also ask someone out on a date for the first time, and I would be told no, because I was a freak.

These experiences influenced my development, especially my self-esteem and sense of identity (i.e. self-concept). As a child, most things are seen as black and white, understanding the abstract isn't a strong ability yet. Viewing my sense of self and my body as distinct and separable was not possible. Thus, I remember struggling and constantly wrestling

with the same questions, “If my body’s not mine then… what am I, Who am I, What did I do wrong, and Why me?” Carrying out this research was rewarding because it highlights areas which I know today are invaluable, such as early identification and intervention. Although I struggled with my own TS, to some degree I knew what I was struggling with. I cannot imagine what it must be like for children and families who don’t have access to specialist care. To *know* something is off, and to go years without a diagnosis which will at least, to some degree, tell them the *what* and *why* of their experience.

School especially was hard for me, I also had my tics mimicked by peers, I had a teacher stack tri-fold presentation boards around my desk so I wouldn’t further disrupt my classmates; who in fact were laughing *at* me, not *with* me. To contribute evidence which may promote positive change for today’s CYP with TD is what makes this project so fulfilling for me. However, my personal connection to the subject matter is also what made carrying out this research project so challenging. Throughout the process, I had an intense fear of letting “my people” down. This led to perfectionistic thinking, heightened anxiety and stress, and of course my tics became worse. Throughout this process, I was reminded of the faces and voices of the many professionals I interacted with over the course of my life, some were substantially influential some less so. I realise now, that over the course of this project and the escalation of my tics, I was an effective self-advocate. I applied the behavioural techniques taught to me when I was young and communicated my needs effectively with the systems that surround me: my training programme, healthcare professionals, colleagues, friends and family. These insights remind me of how influential systems can be, not in becoming self-reliant but learning to know myself and how to authentically engage with the world around me. I am reminded of a quote, which I believe captures the essence of this research project and serves as a fitting way to conclude this paper:

“Let us put our minds together and see what life we can make for our children.”

- Sitting Bull

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Word Count

Thesis section	Excluding references, tables, appendices etc	Inclusive
Thesis summary	273	273
Scoping Review	5733	8412
Empirical Study	3955	5551
Contributions Chapter with Reflective Commentary	2299	3219
Title pages, acknowledgements, abbreviations, contents, word count	n/a	938
Total word count	**12260**	**18393**