Group work for children with Tourette syndrome: A pilot randomised controlled trial evaluating tic severity and neuropsychological outcomes

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Thesis declaration form

I confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

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Overview

This thesis consists of three major components: A literature review, an empirical paper and a critical appraisal.

The literature review outlines a wide-ranging systematic search, describing and evaluating the effectiveness of psychosocial group-delivered treatments for children aged 5 – 18 years with ADHD. Studies included child-only groups as well as those that involved parents, multimodal treatment, or a comparison of these different approaches. Twenty-two studies were described, in which cognitive-behavioural approaches dominated. The methodological quality of nineteen studies was assessed, finding that child-only groups and dual interventions had the poorest methodological quality. Treatment effectiveness findings for ten of these studies were mixed, providing most support for multicomponent approaches.

The empirical paper reports the findings of a pilot randomised controlled trial, which aimed to evaluate tic severity and neuropsychological outcomes following group work for children with Tourette syndrome. The two group-delivered interventions were a Comprehensive Behavioural Intervention for Tics (CBIT) compared with a psycho-educational group. Preliminary evidence indicated both interventions to be feasible and effective, in terms of improving tic severity and inhibitory processes of neuropsychological functioning. CBIT was superior in reducing motor tic severity. Study design, recruitment, testing and data entry was carried out jointly with Rachel Yates, a Trainee Clinical Psychologist from Royal Holloway, University of London.

Finally, the critical appraisal offers reflections on the challenges encountered throughout the process of conducting a moderately-sized pilot RCT, from the early design and recruitment stages through to data collection and analysis. This is combined with a further discussion of the strengths and weaknesses of this study.

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Part 1: Literature Review

Group-delivered psychosocial interventions for children aged 5 to 18 years with Attention-Deficit Hyperactivity Disorder (ADHD): A systematic review

Abstract

Aims

This review aimed to describe and evaluate the effectiveness of psychosocial group-delivered treatments for children aged 5 – 18 years with ADHD.

Method

A wide-ranging systematic search was conducted, combining database searching (PsycINFO, MEDLINE, EMBASE) with reference lists from previous high-quality reviews to cover studies using a range of research designs from 1946 to 2014. Exclusion criteria included social skills and summer treatment programs. Extracted data included: Descriptive information, methodological quality assessment and a narrative synthesis of outcome data.

Results

This review identified 22 group-delivered psychosocial treatments for children with ADHD, which focused on improving ADHD symptoms and associated difficulties. The interventions were primarily based on cognitive-behavioural techniques and varied in their approach (e.g. problem-solving, mindfulness, anger management). Studies included child-only groups as well as those that involved parents (“dual”) or were part of a multimodal treatment package. The methodological quality of 19 studies was analysed. Ten were subsequently included in evaluations of treatment effectiveness. Most evidence was available for groups embedded within a multicomponent approach, with indications of both short and longer-term benefits.

Conclusions

Psychosocial groups, in outpatient clinical and educational settings, are a feasible and acceptable approach for many children with ADHD and their families. Cognitive-behavioural approaches are typically provided. This review supports the use of group

approaches, particularly when embedded within multicomponent treatment packages. Implications for clinical practice and future research are discussed.

Introduction

Background

Attention-Deficit Hyperactivity Disorder (ADHD) is a pervasive and impairing neurodevelopmental disorder beginning in childhood. ADHD symptoms are currently characterised by the Diagnostic and Statistical Manual of Mental Disorders (5th Ed., DSM-5; American Psychiatric Association [APA], 2013), as clustering into 2 areas: Hyperactivity/impulsivity and inattention. ADHD subtypes are defined as predominantly inattentive (ADHD-I), predominantly hyperactive-impulsive (ADHD-H/I) or a combination of these (ADHD-Comb). The International Classification of Diseases (10th Ed., ICD-10; World Health Organization, 1992), uses more restrictive criteria for what is termed Hyperkinetic Disorder (with or without conduct disorder), approximating a more severe combined subtype of ADHD. ADHD has evolved conceptually over the last several decades, being assigned various terms including Hyperkinetic Syndrome (ICD-9; World Health Organization, 1977) and Attention Deficit Disorder with Hyperactivity (ADDH) or without hyperactivity (ADD) (DSM-III; APA, 1980). These varied in terms of emphasis on subtypes of ADHD versus a uni-dimensional conceptualisation. This report will use “ADHD” as an umbrella term to refer to the various conceptualisations of the disorder. Although studies on the neurobiology of ADHD have indicated roles for epigenetic (e.g. Mill & Petronis, 2008) and genetic (e.g. Faraone & Biederman, 1998) factors, it continues to be defined by behavioural symptomatology given that it does not indicate a neurological disease (National Institute for Health and Clinical Excellence [NICE], 2008).

In the UK, prevalence of ADHD has been conservatively estimated as 3.62% for male and 0.85% for female children aged 5-15 years old (Ford, Goodman & Meltzer, 2003). ADHD is likely to affect around 210,000 children aged 5 – 18 years old in England and Wales (NICE, 2008). The estimated worldwide prevalence is 5.29% (Polanczyk et al., 2007). Children with ADHD are at high risk for co-morbidities including low

mood, conduct disorder, learning and social communication difficulties, poor motor control and anxiety disorders (NICE, 2008). In the long-term, ADHD is associated with high risk of delinquency and substance abuse in adolescence and adulthood (Barkley, 2006), educational failure, relationship difficulties and mental illness (Biederman et al., 2006). ADHD is one of the most prominent disorders encountered in paediatric primary care and school contexts. It is associated with extensive costs to juvenile justice systems, health, social care and education services (Telford et al., 2013; Pelham, Foster & Robb, 2007).

Treatment approaches

A variety of treatment approaches have been developed for children with ADHD, with the aim of alleviating symptoms or associated difficulties. Many previous reviews and meta-analyses have focused on evaluating the evidence for particular approaches, such as pharmacological treatments (e.g. Swanson et al., 1993) or specific psychosocial interventions (e.g. Storebo et al., 2011, social skills; Conway, 2012, psychodynamic therapy). The term “psychosocial” tends to refer to non-pharmacological interventions, emphasising psychological or social factors over biological approaches. Some previous reviews have looked more generally at all psychosocial approaches (e.g. Pelham & Fabiano, 2008; Chronis, Jones & Raggi, 2006). Others have more widely evaluated the effectiveness of both pharmacological and psychosocial treatments (e.g. Van der Oord et al., 2008; Purdie, Hattie & Carroll, 2002). The development of the NICE (2008, updated in 2013) clinical guidelines for managing ADHD constitutes a broad review incorporating all available empirical evidence up to 2007.

While psycho-stimulant medication is widely-used and efficacious, NICE (2008) now advise that this should be reserved for children with severe symptoms or impairments only. This is because there are many limitations to pharmacological treatment, which can include: Patient choice (families often have concerns or moral/ethical objections),

non-response, limited clinical benefits of responders (e.g. less impact on functional impairments), intolerable side effects and poor evidence for long-term effects. In addition, medication does not address commonly impairing co-morbid difficulties. This guidance is given within the context of an emerging evidence base for psychological therapies. For children 5 – 18 years old with moderate symptom severity, first-line recommended treatments by NICE (2008) are group parent-training and education (e.g. ADHD psycho-education, behaviour management techniques, improving communication), either alone or in combination with group-delivered psychological treatment (cognitive-behavioural therapy, CBT; and/or social skills training). The latter is emphasised particularly for older children and adolescents. This is in contrast to recommendations for pre-school children (under five years), which are based on parent interventions only. A recent Cochrane review on social skills groups for adolescents with ADHD by Storebo and colleagues (2011) found limited high-quality evidence to recommend or refute this approach.

The NICE (2008) recommendation of child-focused group CBT interventions is fairly broad. In the guidelines, CBT is described as a psychological treatment which encourages clients to recognise the links between thoughts (or cognitions), feelings and behaviours, and change unhelpful or maladaptive processes into more beneficial outcomes. CBT by definition combines both cognitive and behavioural approaches, although in clinical practice is typically weighted towards one or the other. In ADHD treatments, cognitive therapy can involve a variety of approaches. Self-instructional training is frequently implemented, aiming to teach children to cultivate a more strategic, systematic and self-evaluative thinking style when confronted with tasks or problems by developing schemas and step-by-step decision-making processes (NICE, 2008). Other approaches such as cognitive (e.g. working memory, attentional) training are receiving emerging empirical support (Toplak et al., 2008), however are not yet considered evidence-based treatments. Krisanaprakornkit and colleagues

(2010) conducted a Cochrane review on meditation therapies for ADHD but found very limited evidence, stating that more trials are needed.

Behavioural approaches typically involve the 'use of rewards or reinforcers that are judged likely to encourage the young person to implement targeted changes in motor, impulse or attentional control' (NICE, 2008, pp.151). This typically involves token reward systems, in which daily or weekly rewards are earned by individuals (or the group) to facilitate motivation and reinforce desired behaviours. To discourage undesirable behaviours (e.g. disruptiveness), several techniques are often implemented such as response cost procedures (loss of a reinforcer e.g. tokens or rewards removed) and time outs. These approaches are often referred to as behaviour (or contingency) management techniques. Behavioural interventions also often involve changing aspects of the environment that precede or follow undesired behaviours (Chronis, Jones & Raggi, 2006). There is strong evidence that behavioural treatments for children with ADHD are highly efficacious (Fabiano et al., 2009). After conducting extensive reviews, Pelham and Fabiano (2008) concluded that behavioural parent training, behavioural classroom management (i.e. applying contingency management techniques in a classroom context) and intensive peer-focused behavioural interventions implemented in recreational settings (e.g. summer programs, often focused on social skills) can be considered evidence-based.

The NICE (2008) recommendation for child-focused group CBT interventions promotes the use of behaviour management techniques and advises clinicians to focus CBT programs on areas such as oppositional behaviour, problem-solving, self-control, listening skills, assertiveness, self-esteem and emotional regulation. Structured programs using active learning methods (e.g. therapist modelling, role play) and individualised components (e.g. homework) are advised. These guidelines are useful in summarising best practice evidence but still have significant limitations. The NICE guideline search strategies are limited to randomised controlled trials

(RCTs) only, of which there were none involving adolescent (13+) populations and very few focusing directly on group psychological therapies. It was concluded that more evidence on the effectiveness of psychological interventions for school-aged children with ADHD, particularly addressing these aforementioned gaps, was needed. Group CBT recommendations in NICE (2008) appear to be based primarily on cost-effectiveness analyses in addition to a limited number (*n* = 10) of group studies (excluding social skills groups): Six on the Multimodal Treatment Study of Children with ADHD (MTA; e.g. MTA Cooperative Group, 1999), three on other multimodal psychosocial or CBT approaches (Abikoff et al., 2004a & 2004b; Bloomquist, August & Ostrander, 1991) and one on a stress-management group (Gonzalez & Sellers, 2002). The MTA is one of the largest and most frequently cited studies, with highly acclaimed methodology. The term “multimodal” or “multicomponent” are commonly used to describe when several interventions are delivered in parallel, for example, pharmacological and psychosocial treatments (for children, parents and teachers). The MTA study (MTA Cooperative Group, 1999) evaluated best practice for medication management (MedMgt) and intensive multimodal behaviour therapy (MBT; child, parent, teacher), alone or combined, in a summer treatment program setting (with a routine community care control group, CC). After 14 months of treatment it was found that MedMgt, alone or combined conditions, was superior to MBT and CC in reducing ADHD symptoms, although slight advantages were suggested for the combined treatment in terms of other functional measures (e.g. social skills, academics, internalising and externalising problems). Children with parent-reported anxiety and disruptive disorder problems were also found to respond better to MBT and combined treatments. At 3 year follow-up (Jensen et al., 2007) there were no significant differences between treatments, with all children showing symptom improvement from baseline.

Van der Oord and colleagues (2008) conducted a meta-analysis of the efficacy of methylphenidate, psychosocial treatments and their combination in school-aged

children with ADHD. This presented similar findings to the short-term effects observed in the MTA study, with benefits of pharmacological and psychosocial treatments being indicated for different types of outcomes. While medication was superior (large effect sizes) in reducing ADHD symptoms, both treatments were equally effective (medium effect sizes) in terms of functional outcomes (social behaviour).

For all ADHD severities, NICE (2008) recommends integrated care including psycho-education and psychological approaches (including parent and teacher involvement), given the typically wide range of impairments and co-morbidities experienced. Kaiser, Hoza and Hurt (2008) state that evidence for stand-alone psychological treatments is limited and these are most effective when delivered within a multimodal approach. It was advised that more attention should be paid towards evaluating and comparing different levels of approaches (e.g. stand-alone, combined or multimodal treatments) in order to identify active treatment components.

Rationale

There are no known published systematic reviews to date that have focused on group-delivered, child-focused psychosocial treatments for children aged 5 – 18 years with ADHD. NICE (2008) recommends this approach however bases this on limited evidence given the lack of high-quality trials conducted, particularly for adolescent populations. It is vital to understand and collate the evidence for groups more directly, explaining more clearly how these might be delivered and evaluating how effective they are. RCTs are considered the gold-standard for establishing treatment efficacy and further studies may have been conducted since the NICE (2008) guidelines were published. However, there is also clearly the need to examine evidence from a wider range of study designs. An all-inclusive approach in the context of limited evidence is recommended when conducting systematic reviews of interventions (Jackson & Waters, 2005; Fabiano et al., 2009).

Objectives

The aims of this review are as follows:

Objective 1: Description of groups

What types of group-delivered psychosocial interventions for children and adolescents with ADHD are described in peer-reviewed studies over the last several decades?

- What settings are they in?
- Who are they facilitated by?

Objective 2: Effectiveness of groups

a) What is the methodological quality of the selected studies?

b) How effective are these interventions, and in what ways? What are the limitations?

c) Is there overall support for using that group approach with this population?

Method

A systematic search was conducted, based on standard mental health related bibliographic databases. The aims of the review dictated the approach taken. It was important to have a broad enough scope (low specificity) to facilitate the selection of studies with a range of research designs and aims, whilst having high sensitivity to detect the most appropriate studies given the limited resources of this review. The term “group” is commonly used to describe treatment conditions and not necessarily group-delivered interventions, thus a large number of studies were expected in response to the search terms.

Eligibility criteria

This review aimed to identify studies with specific characteristics, as outlined in Table 1.

Table 1: Desired study characteristics

Population of interest	Children and adolescents (5 – 18 years old) with ADHD
Interventions	Child-focused, psychosocial group-delivered interventions (alone or in combination with other parallel interventions delivered separately or jointly with the children e.g. parent, school, medication)
Comparator	Healthy or clinical control groups of all types (e.g. waitlist, treatment as usual, other active intervention), desirable but not essential for inclusion
Outcomes	Any outcomes relating to change associated with child groups, with a focus on child outcomes but consideration of other areas likely to be related (e.g. reduction in parental stress)
Study design	All accepted except case series (e.g. RCTs, controlled trials, cohort studies)

Inclusion and exclusion criteria

This review focused on studies involving young people aged 5 – 18 years with a diagnosis of ADHD (any diagnosis or subtype; e.g. ADHD-I or -H/I or –Comb; ADD, ADDH, Hyperkinetic Disorder) or identified hyperactivity of a clinical level. Wide-ranging methods of confirming ADHD diagnosis in participants were considered acceptable, including previous diagnoses from other clinicians and the use of recognised rating scales and / or diagnostic interviews. Worldwide peer-reviewed published journal articles, written in English, were eligible for inclusion. There was no scope for the searching of grey literature. Many interventions for young people with ADHD typically include the involvement of parents or teachers, thus combined or multicomponent treatment packages delivered in parallel were included if they involved a child-focused group-delivered psychosocial intervention. Medication status of participants did not affect eligibility. Psychosocial interventions provided as an adjunct to pharmacological treatment were accepted. Groups could be conducted in various treatment settings (e.g. clinics, schools, laboratory classrooms), however

were required to be outpatient or community-based. Articles were accepted from any date of publication.

Exclusion criteria were as follows, being primarily due to limited resources unless otherwise stated:

- Studies focusing explicitly on ADHD in combination with one or more co-morbid diagnosable disorders (e.g. “aggressiveness” could be included, but not Autistic Spectrum Disorder, etc). General reporting of co-morbidities in a sample for descriptive purposes was acceptable.

- Child group interventions with the sole aim of social skills training (given an extensive previous Cochrane review by Storebo et al., 2011).

- Group treatments considered less relevant (e.g. yoga, music / art therapy, physical training, groups purely based only on behaviour / contingency management or academic skills training).

- Approaches based more on biological or neuropsychological models (e.g. neurofeedback, cognitive training).

- Summer treatment program settings (e.g. MTA Cooperative Group, 1999): This treatment context was considered unique compared to others and difficult to compare. It is also resource-heavy and would be unrealistic for many services and clinicians to implement.

Some studies were included for descriptive purposes but excluded in effectiveness analyses (e.g. studies including both adolescents and adults). All exclusions were carefully made following a pre-devised structured process (see Appendix 1).

Information sources and search strategy

Three electronic databases were searched to identify relevant studies:

1. PsycINFO: 1st January 2007 – 17th May 2014
2. MEDLINE: 1st January 2007 – 17th May 2014
3. EMBASE: 1st January 2007 – 17th May 2014

Pre-defined search strategies were thoroughly trialled prior to the search to ensure that all relevant terms were included, also informed by the search strings of previous high-quality systematic reviews on ADHD and group therapies. The search strategy for each database included subject heading (database-specific) and multi-purpose (.mp; title, abstract, key concepts and more) searches. Search terms were based around concepts of ADHD, young people, groups and interventions. Limitations applied were: Publication date (as above), peer-reviewed journal articles, English, humans and age (roughly 0 – 18 years; limits available varied between databases, see Appendix 2). Limits were necessary due to the large number of initial hits (7000+). Publication date limits (Jan 2007 to present) were based on time periods not well-covered by previous reviews (see below). The complete search strategy, with Boolean operators, for each database is outlined in Appendix 2.

Previous reviews and other sources

The reference lists of several key reviews were thoroughly scanned to identify additional papers. This is typically done when searches produce over 5000 initial hits (NICE, 2008). Appropriate previous reviews were primarily identified through The Cochrane Database of Systematic Reviews (Issue 5; Inception to 17th May 2014) and the University of York Centre for Reviews and Dissemination databases (accessed on 17th May 2014).

Reference lists from the following reviews were combined to identify studies of various research designs and treatment approaches, as most were biased towards specific types:

- NICE ADHD guidelines (2008 – authors searched databases from inception to 18th December 2007; 2013 brief update but not full search): RCTs only
- Conway et al. (2012 – dates authors searched not reported, but selected studies in the results were published from 1994 to 2011): Psychodynamic therapies only
- Krisanaprakornkit et al. (2010 – authors searched from inception to January 2010): Cochrane review of meditation therapies for ADHD, variety of study designs
- Fabiano et al. (2009 – authors searched from inception to December 2006): Meta-analysis of all research design types but limited to behavioural therapies
- Toplak et al. (2008 – authors searched from March 1981 to May 2007): Systematic review of cognitive and cognitive-behavioural treatments for ADHD

No previous reviews were identified that evaluated treatment outcomes for all types of psychosocial therapies of all study designs.

Titles and abstracts of all identified papers were initially scanned. Following this, full-texts were inspected to ensure eligibility. Additional papers were identified through searching the reference lists of included studies. A key informant in the field of paediatric clinical psychology was also consulted.

Methodological quality assessment

Numerous tools are available for use in evaluating the methodological quality or risk of bias in healthcare or clinical effectiveness studies. For the present review, a tool appropriate for quantitative studies was required, assessing a range of randomised and non-randomised research designs. The Effective Public Health Practice Project (EPHPP) was selected. This domain-based assessment tool has been used to critically appraise research evidence since 1999. It has been judged appropriate and

recommended for use with systematic reviews of effectiveness (Deeks et al., 2003; Jackson & Waters, 2005) and has acceptable inter-rater reliability, as well as good content and construct validity (Thomas et al., 2004). Alternative tools were considered, such as the checklist for randomised and non-randomised studies (Downs & Black, 1998), however the Cochrane Collaboration (Higgins & Green, 2011) recommends domain-based assessments rather than checklists or scales.

The EPHPP is comprised of six main component sections which contribute towards a global rating: Selection bias, study design, confounders, blinding, data collection methods and withdrawals or drop-outs. Two additional sections on intervention integrity and statistical analysis are included descriptively but do not contribute to the global rating. Each of the main component sections are given a rating of 'Strong', 'Moderate' or 'Weak' based on standardised guidelines and a scoring dictionary (see Appendix 3 for a summary table of how each section is scored). These ratings are combined to give studies a global rating of Strong (no Weak component ratings), Moderate (no more than one Weak rating) or Weak (two or more Weak ratings) methodological quality.

Data extraction

The author carried out the study selection process independently. Data extracted varied according to the different aims of the review and included descriptive, methodological quality and outcome data. These are more fully outlined in the Results.

Synthesis of results

Studies were considered too heterogeneous in their aims, dependent measures, sample populations (i.e. ADHD subtypes) and interventions to support a meta-analytic approach. Results were instead synthesised following a narrative approach, incorporating measures of effect size descriptively when reported.

Results

Study selection

The systematic search strategy employed (combining database and reference list searching) led to the initial identification of 6754 studies, the majority of which were identified by database searches. A total of 4567 studies were retained following the removal of duplicates.

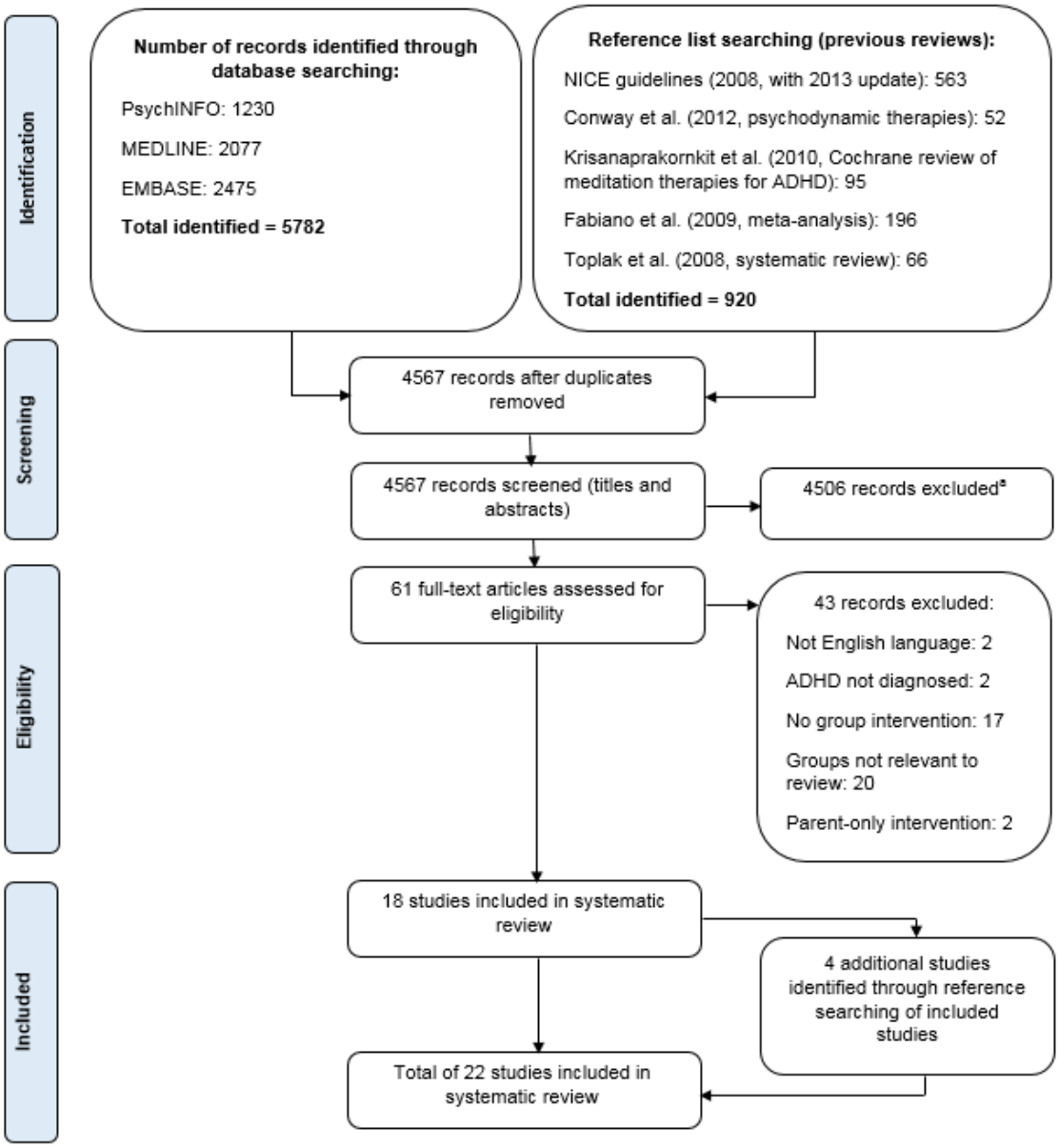
Titles and abstracts were screened, leading to the exclusion of 4506 studies. Exclusions were carefully made following a pre-devised, structured process (see Appendix 1). 61 full-text articles were then assessed for eligibility. 43 of these were excluded for reasons including: Not involving a group treatment (*n* = 17), having an irrelevant or non-psychosocial group approach (*n* = 20, including groups focusing on social skills or academic skills only), full article not written in English (*n* = 2), no clinical diagnosis/level of ADHD or hyperactivity (*n* = 1; only traits of inattention or impulsivity) and for involving parent groups only (*n* = 2). Next, the reference lists of the remaining 18 eligible articles were examined to identify any additional relevant studies. Four were deemed eligible. Papers recommended by the key informant had already been identified.

A total of 22 studies were classified as eligible to be included in this review. The complete study selection process is illustrated in Figure 1.

Objective 1: Description of groups

The study characteristics of the 22 identified studies are presented in Table 2. Data extracted included sample size and characteristics, study location (globally), exclusion criteria, method of verifying ADHD diagnosis, group setting, profession of group facilitators (“delivery”), intervention summary and group details.

*Figure 1: Study selection process**



** Figure based on PRISMA guidelines (Moher et al., 2009)*
[a] Exclusions were made following a pre-defined structured process (see Appendix 1)

Table 2: Key characteristics of selected studies (n = 22)

Primary author (date)	*n*	Sample characteristics	Study location	Exclusion criteria	ADHD assessment	Group setting and delivery	Interventions	Group size, duration and details*
Barkley (1980)	6	Age range 7-10yrs; All males	US	IQ<80; Serious neurological or psychiatric problems	*Teachers:* Conners Teacher's Questionnaire, >2SDs above mean *Parents:* 1. Modified Werry-Weiss-Peters Activity Rating Scale, >2 SDs above mean 2. Parental interview	Experimental classroom (after-school); Delivery unclear	**Self-control training**; Groups: Free play, 20mins large group activity (academic / social skills topics), 45mins small group (continuing same topic) or individual work (incorporating self-monitoring: signal, child recorded if on/off task, rewarded for accuracy), free play. **Week 1:** Topics presented in lecture / discussion format (BASELINE) **Weeks 2-4:** Topics presented using 4-step problem-solving / self-instructional techniques (PHASE A) **Week 5:** Lecture/discussion format (REVERSAL) **Week 6-8:** Problem-solving / self-instructional techniques implemented again (PHASE B)	Groups of 6; 8 weeks (32 sessions; 4 x 2hrs weekly); Mixture of group and individual work
Gonzalez (2002)	42	Aged 9-12; 79% males, 21% females; Average to above average intelligence; 38% White, 24% Hispanic, 17% African American, 17% mixed race and 4% Asian	US	No other diagnosed medical, psychiatric, or neurological problems	Previous diagnosis of ADHD was accepted	Outpatient clinic; Clinical social worker experienced in child therapy	**1. Therapist-led child stress-management group:** Psycho-education, breathing, muscle relaxation, imagery, exercise awareness and physical fitness, time as a stressor, assertiveness, handling anger, expressing emotions and making friends. **2. Stress-management techniques taught by parents** to their children (1:1 at home) **3. Control group (no intervention)**	Size unclear ("small groups"); 4 weeks (8 sessions; 2x30-40mins weekly); Parents not involved in therapist-led group
Hinshaw (1984) - Study 1 only	21	Aged 8-13; All males; All hyperactive; Average or above average intellectual functioning	US	Mental retardation, severe emotional disturbance, or organicity; Acute family distress; Known poor reaction to methylphenidate; Methylphenidate 3 months prior to the study; Prescribed other psychotropic medication	Primary diagnosis of hyperactivity or hyperkinesis by the child's physician	Outpatient clinic (university-based); Clinical psychology graduates	**1. Small group training in cognitive self-regulation skills + methylphenidate:** Self-instructional and problem-solving cognitive-behavioural techniques for first 4 sessions, practiced in academic/fine motor tasks and interpersonal situations. Session 5: Behavioural peer (verbal) provocation test to assess ability to control reaction. **2. Small group training in cognitive self-regulation skills + placebo:** Same as condition 1 with placebo medication for the 3-week period.	Groups of 3; 3 weeks (6 sessions; 2x2hrs weekly)

Looyeh (2012)	14	Aged 9-11; All females	Iran	No prior history of treatment for ADHD	Child Symptom Inventory (CSI) for ADHD diagnosis; DSM-IV diagnostic evaluations to differentiate possible behavioural or emotional disorders	School setting (after hours); School psychologist	**1. Group narrative therapy:** Aim to communicate problem-saturated narratives and construct more positive narratives. Stories were told and acted out (by facilitators and children, using many play therapy techniques), each time with a different focus (e.g. emotion recognition, techniques to manage hyperactivity and enhance self-control, alternative behaviours, verbalizing consequences and so on). **2. Waitlist control**	Groups of 3-4; 6 weeks (12 sessions; 60mins x2 weekly)
Miranda (2000)	32	Aged 9-12; 78% males, 22% females; All Caucasian; 50% co-morbid aggression and ADHD, 50% no aggression	Spain	IQ<80; Psychosis; Severe neurological, sensory, or motor impairment; Score < 7 on the Aggression (ODD) factor in the Iowa Scale for Teachers (aggressive condition); Children currently on medication for ADHD	1. Abbreviated Conners Rating Scale for Teachers (score >15) 2. DSM-III-R parent interview (score >16 on ADHD section), chronic duration and early onset criteria	School setting (after-hours); PhD-level psychologists	Evaluating effectiveness of two groups for ADHD vs ADHD + aggression (each group included both types) **1. Cognitive-behavioral self-control training:** Trained to use self-instructional strategies when problem-solving (applied to interpersonal situations, emotion recognition and individual targeted behaviour problems). Also behavioral contingencies e.g. token system, social reinforcement. **2. Combined treatment: Cognitive-behavioral self-control training (as above) + anger management training:** Sessions 1-15 of self-control training, with anger management adaptations to final 7 sessions (triggers, relaxation, coping strategies, etc).	Groups of 4; 12 weeks (22 sessions; 60mins x2 weekly)
Ozcan (2013)	33	Aged 6-11; 91% males, 9% females; All Caucasian	Turkey	History of head trauma or neurological illness, developmental delay or other axis I psychiatric disorders (except ODD); Change in medication during the study (if prescribed)	DSM-IV-TR criteria for ADHD diagnosis. To exclude other psychiatric disorders: The Children's Depression Inventory / State-Trait Anxiety Inventory / Learning Disorders Checklist	School setting; "Primary researcher" trained in delivering the program	**''I Can Problem Solve" (ICPS) program**: Children with ADHD were taught and encouraged to practice interpersonal cognitive problem-solving strategies. They were encouraged to evaluate their own thoughts, think of solutions to a problem, consider consequences and modify their behaviour. There was a focus on social difficulties, prevention of high risk behaviors and emotion recognition. The program was adapted to be delivered in Turkish.	Groups of 7-9; 14 weeks (session number unclear; Roughly 30mins 2x weekly)

Zylowska (2008)	32	Aged 15 or older; 25% adolescents, 75% adults; 38% males, 62% females; 53% ADHD-Comb, 41% ADHD-I, 6% ADHD-H/I; Lifetime co-morbidity: 78% any mood disorder, 34% any anxiety disorder, 25% ODD	US	Substance dependence within past 6 months; History of psychotic illness; Bipolar I disorder; Mental retardation; Borderline /antisocial personality disorder; CD; Chronic suicidal or self-injurious behavior	ADHD (DSM-IV criteria), verified by: Schedule for Affective Disorders and Schizophrenia for School-Age Children–Present and Lifetime Version. Self-report and observer behavioral ratings from ADHD-IV scale (adults) or SNAP-IV (adolescents)	Outpatient clinic; Experienced mindfulness instructor and ADHD researchers	**Mindful Awareness Practices (MAPs) for ADHD program:** Mindfulness meditation training adapted for adults and adolescents with ADHD (e.g. psycho-education on ADHD, shorter sitting meditation periods, emphasising mindful awareness in daily life and using visual aids to explain concepts).	Groups of 6-8; 8 weeks (8 sessions, 2.5hrs weekly)
Fields (2011)	40	Aged 7-12; 80% males, 20% females; Ethnic backgrounds: 75% White, 15% African American, 2.5% Latino, 7.5% Asian; Roughly half having ongoing treatment with other professionals	US	Co-morbid diagnosis of psychotic disorder	Any previous diagnosis of ADHD was accepted (any of the 3 subtypes)	Outpatient family medicine clinic; Experienced psychologist and assistant psychologist	**1. Psycho-educational child and parent groups:** Sessions covered: What is ADHD, communication skills, behavioural management, problem solving and family anger management. **2. Waitlist control group**	Groups of 3-5; 5 weeks (5 sessions; 1hr weekly); Separate child and parent groups
Sibley (2014)	28	Aged 14-18; 70% males, 30% females; 87% White (predominantly Hispanic), 9% mixed, 4% Black; Co-morbidities: 35% ODD, 9% CD	US	IQ<80; History of an autism spectrum or psychotic disorder	1. *DSM-IV-TR* criteria for ADHD (phone screening) 2. Parent semi-structured *diagnostic interview* with probes for symptom severity and situational variability (combined with core teacher ratings). Diagnosis agreed by two PhD-level psychologists.	Setting unclear; Clinical psychology doctoral students / counseling master's student / undergraduate assistant	**1. Supporting Teens' Academic Needs Daily-Group (STAND-G) - Adolescent group behavioural intervention + parenting skills group**: *Teen group:* Psycho-education and modules in academic (study skills, goal-setting), organizational (use of planners, daily routines) and communication (problem-solving, negotiation) skills. Token/reward system. *Parent group:* Taught to monitor academics, build in routines and apply behavioral principles to support academic performance. **2. Waitlist control group**	Groups of 5-10; 8 weeks (8 sessions, 90mins weekly); Parent and teen groups separate but joined for first 10mins and final 20mins

Van der Oord (2012)	22	Aged 8-12; 73% males, 27% females; 55% ADHD-Comb, 31% ADHD-I, 14% ADHD-H/I; 14% co-morbid ODD; Previous treatment: 32% prior mental health treatment for behaviour problems, 14% neurofeedback, 14% parent training	The Netherlands	IQ<80; Inadequate mastery of Dutch by child or parents; Severe behavioral problems (i.e. CD diagnosis on the ADISC); Co-morbid developmental disorder (e.g. autistic); Family participating in another ongoing psychological intervention.	DSM-IV diagnosis of ADHD established with the parent and child version of the Anxiety Disorder Interview Schedule for Children (ADISC; ADHD, ODD and CD sections)	Outpatient clinic; Experienced cognitive-behavior therapists / mindfulness trainers	**1. Mindfulness training - Child group + mindful parenting training:** Based on Mindfulness-Based Cognitive Therapy and Mindfulness-Based Stress Reduction Training, adapted for children with ADHD and their parents (e.g. highly structured, visual aids, minimal distractors in environment, applying mindfulness in difficult situations). Child group included a token reward system. **2. Waitlist control** (same children, pre-intervention)	Groups of 4-6; 8 weeks (8 sessions, 90mins weekly); Part of the 1st, 6th and the whole last session were joint, with parents and children together (otherwise separate groups)
Van de Weijer-Bergsma (2012)	10	Aged 11-15; 50% males, 50% females; 50% ADHD-Comb, 40% ADHD-I, 10% ADHD-H/I	The Netherlands	Commencement of medication during the treatment or follow-up period	Not reported	Outpatient clinic; Experienced cognitive-behavior therapists / mindfulness trainers	**Mindfulness training - Child group + mindful parenting training:** Adapted for ADHD (e.g. stimulus-free environment, structured, visual aids). Adolescents learnt to focus and enhance their attention, awareness and self-control through mindfulness training (emphasis on applying this in stressful situations e.g. distractibility at school, conflict with others). Token/reward system. Parent group targeted common difficulties/conflicts of parenting children with ADHD, and (potentially) parents' own ADHD symptoms.	Groups of 4-6; 8 weeks (8 sessions, 90mins weekly); Separate teen and parent groups, except joint booster session 8 weeks after final group

Bloomquist (1991)	52	Mean age of 8-9yrs; 69% males, 31% females; Mild-moderately severe ADHD; 35% met criteria for ODD	US	Mental retardation; epilepsy; severe emotional disorder; pervasive developmental disorder	*Teacher:* Initial school-wide screening, inattentive factor in Child Behavior Checklist-Teacher Rating Form (T score >60) *Parent:* Child Behavior Checklist, parent version (T score >65); Diagnostic Interview for Children and Adolescents - Revised, ADHD diagnosis	School; Educational psychologists & undergraduate psychology students	**1. Multicomponent Cognitive-Behavioral Therapy (MCBT)**: Child, parent, and teacher (school-wide) training. Children: Learnt problem-solving steps, applied in specific areas (e.g. friendships, anger management). Token reward / response cost procedures. Teachers: see below. Parents: Psycho-education, problem-solving techniques and behaviour management approaches. **2. 10-week Teacher only:** 10 weeks; All teachers in school invited to one 2hr and six 45- to 60-min consultations. Psycho-education, problem-solving and behavior management methods. **3. Waitlist control**	Groups of 6-8; 10 weeks (20 sessions; 2x1hr weekly); Separate child and parent groups
Horn (1991)	128	Aged 7-11; 77% males, 23% females; 8% co-morbid CD; 15% co-morbid ODD; 85% White, 9% African American, 4% Hispanic, 2% Asian American	US	Severe physical impairments; Intellectual deficits; Psychosis in child or parent(s); Co-morbid anxiety or depression; Child currently taking medication for ADHD, not willing to discontinue it for the duration of the study	DSM-III-R criteria - Clinical interview with parents, CPRS (Score > 2 SDs from mean for Hyperkinesis), CTRS (score > 2 SDs from mean for Hyperkinesis) and a battery of psychometric tests.	Outpatient psychology clinic (university-based); Clinical psychology graduates / undergraduate psychology student	**1. Medication placebo alone** **2/3. Low/High dose stimulant therapy alone** **4. Medication placebo + behavioural parent training + child self-control group + school consultation:** Child: 6-step problem-solving technique, with deep muscle relaxation. Parents: Behaviour management, bibliotherapy, individualised behaviour projects with child. School: Teachers contacted to learn/reinforce skills from child groups. Daily home report card. **5/6. Low/High dose stimulant therapy + behavioural parent training + child self-control group + school consultation** (see above) **7. Control group** (non-ADHD, no intervention)	Groups of 6 (slightly more in parent groups); 12 weeks (12 sessions, 90mins weekly); Separate child and parent groups
Ialongo (1993) - *9 month follow up of Horn (1991)*	117	*See Horn (1991)*	US	*See Horn (1991)*	*See Horn (1991)*	*See Horn (1991)*	*See Horn (1991)*	*See Horn (1991)*

Miranda (2011)	42	Aged 7-10; 85% males, 15% females; All Caucasian	Spain	Psychosis; Neurological damage; Sensory or motor deficits; Vision or hearing difficulties; IQ<80; Child currently on medication for control of ADHD symptoms	Parent and teacher interviews to ensure DSM-IV-TR ADHD criteria met	School setting (after-hours) / community; Specialist therapists	**1. Cognitive-behavioral child group + parent group + teacher training:** *Children* -Self-instruction and problem-solving techniques applied to inhibitory control, anger management and social skills. Token reward system. *Parents* - Psycho-education, behavior modification techniques and various modules (e.g. academic support, emotional regulation, relaxation, problem-solving). *Teachers* - Postgraduate course on behavioural and academic management of ADHD (also classroom adaptations). Daily home-school card. Reinforcement of skills learnt in groups. **2. No treatment control group**	Groups of 4 (parent groups larger, 13-14); 10 weeks (child groups: 16 sessions, 45mins each; Parent groups: 10 sessions, 2hrs weekly); Separate child and parent groups
Pfiffner (2007)	69	Aged 7-11; 67% males, 33% females; All ADHD-I; 51% White, 17% mixed, 16% Asian, 10% Hispanic, 6% African American; Co-morbidities: 23% ODD, 1% Depressive disorder, 12% Anxiety disorder	US	IQ<80; Child not attending school full time; Vision or hearing impairments; Severe language delay; Major neurological illness, psychosis, or pervasive developmental disorder; child in same classroom as another participant or sibling already enrolled	DSM-IV criteria met for ADHD-Inattentive type; Parent interview - Modules (ADHD, ODD/CD, anxiety, mood disorders) from the Schedule for Affective Disorders and Schizophrenia for School-Age Children - Present and Lifetime Version	Outpatient clinic; Clinical psychologist, pre- and post-doctoral psychologists	**1. Child Life and Attention Skills (CLAS) Program - Child skills group training + Parent training + Family sessions + Teacher consultation**: *Child groups:* Independence skills (organization, daily living, study skills, self-care, time management), problem-solving, social skills, specific ADHD-I difficulties (e.g. day-dreaming). Token reward system. *Parents:* Psycho-education (ADHD-I), behaviour management and intervention (e.g. modifying antecedents/ environment). Reinforcing child group skills learnt. *Family sessions:* Individualising programs, reinforcing groups. *Teachers:* Behavioral interventions and classroom adaptations for ADHD-I (initial consultation); 4-5 meetings with family and therapist; Individualised goals. Daily home-school card. Reinforcing groups. **2. Waitlist control group (**1st cohort only) or **Treatment as usual** (2nd to 5th cohorts)	Groups of 6-8 (slightly more in parent); 12 weeks (*children and parents:* 8-10 1.5hr sessions; *family:* 4-5 sessions; *teachers*: 30min consultation; 4-5 family/therapist meetings); Separate groups (joining for 15mins at end). Monthly follow-ups

Pfiffner (2011)	37	Aged 7-11; 76% males, 24% females; 35% Caucasian, 26% mixed, 19% Asian, 14% African-American, 3% American Indian, 3% Latino; Co-morbidites: 46% ODD	US	Significant visual or hearing impairment; Severe language delay; Psychosis; Pervasive developmental disorder; IQ<80	1. *Child Symptom Inventory (CSI)*: 6 or more inattention symptoms and/or 6 or more hyperactive-impulsive symptoms (parent/ teacher-rated) 2. *Impairment Rating Scale*, parent/teacher rated (score >3 in at least one domain)	School setting; Learning Support Professionals (LSPs; masters-level mental health clinicians)	**Collaborative Life Skills Program (CLS) - Child skills group training + Parent training + Teacher consultation/classroom component:** Adapted from the clinic-based CLAS Program (*see Pfiffner, 2007)* for a school context. No family sessions. All components implemented by LSPs. Adaptations to the child and parent groups included slightly shortened session length (with two extra child-parent celebratory sessions) and content appropriate for range of ADHD types (not just ADHD-I). There was also more of a focus on self-control strategies, emotion management and limit setting. Teachers: Shorter initial (group) meeting but similar number of individual meetings with parents/child/teacher/LSP	Groups of 6-7 (slightly more in parent groups); 12 weeks (*Children:* 10 40min sessions + 5 individual with parents/teacher/LSP; *Parents:* 10 1hr sessions; T*eachers:* Up to 6 sessions); Separate parent/child groups
Pfiffner (2013)	60	Aged 7-11; 70% males, 30% females; 40% Caucasian, 21% mixed, 14% Asian or Pacific Islander, 12% African American, 11% Hispanic/Latino, 2% American Indian; 49% co-morbid ODD	US	IQ<80; Significant visual or hearing impairments; Severe language delay; Psychosis; Pervasive developmental disorder; Children in full-day special classrooms	*See Pfiffner (2011)*	*See Pfiffner (2011)*	**Collaborative Life Skills Program (CLS) - Child skills group training + Parent training + Teacher consultation/classroom component:** *See Pfiffner 2011 for details. Same intervention, with aim to extend previous findings by using a larger, complete sample of schools and a broader array of educationally relevant outcomes. Another aim to evaluate potential mechanisms underlying treatment changes.*	Group size unclear; 12 weeks (*see Pfiffner 2011*); Separate child and parent groups

So (2008)	90	Aged 7-9; 86% males, 14% females; All of Chinese ethnicity; All ADHD-Comb (mostly severe); Co-morbidites: 48% ODD, 28% anxiety, 17% dyslexia, 6% depression, 6% CD	Hong Kong	IQ<80; Significant physical disability; Past exposure to stimulant medication (methylphenidate) for more than 2 weeks; Parental intellectual impairment or current psychosis	Diagnosis of ADHD (combined type) according to DSM-IV	Laboratory classroom setting; Child psychiatrists (medication), paraprofessi-onals (nurses, teachers, and OTs); A clinical psychologist	**1. Methylphenidate only:** Optimally titrated **2. Methylphenidate + Behavioural Treatment (BT; child groups + parent training + teacher involvement):** *Child groups:* Training in self-monitoring, problem-solving skills (e.g. academic work, interpersonal conflicts) and anger management. Token reward and response cost procedures. *Parent training:* Behaviour/stress management, reinforcing child group skills. In second half of the program, parents attended as observers then gradually as co-therapists. *Teachers:* 2 phonecalls from group leaders to discuss behaviour management and the home-school report card (with reward system).	Groups of 8-9 (slightly more in parent groups); 6 months (both conditions); *Child groups:* 24 sessions, 100mins weekly; *Parent training:* 24 sessions, 90mins weekly
Van der Oord (2007)	50	Aged 8-12; Mixture of males and females; 62% ADHD-Comb, 32% ADHD-I, 6% ADHD-H/I; 89% Caucasian, 9% mixed, 2% Caribbean; Co-morbidities: 46% ODD, 4% CD	The Netherlands	IQ<75; Inadequate mastery of the Dutch language by child or parents; History of methylphenidate use	DSM-IV diagnosis of ADHD established with the parent version of the *Diagnostic Interview Schedule for children (DISC-IV)*	Outpatient clinic; Child-psychologists (familiar with behavioural treatment of ADHD)	**1. Methylphenidate only:** Optimally titrated **2. Methylphenidate + multimodal behaviour therapy (child cognitive-behavioural group therapy + parent training + teacher training):** *Child groups:* Problem-solving; Relaxation; Academic / interpersonal problems; Token reward system. *Parents: P*sycho-education, structuring the environment, positive attending skills, behaviour management. *Teachers:* 2hr workshop (psycho-education, structuring the classroom for ADHD, contingency management). School-home daily report card (with reward system).	Groups of 4-5 (slightly more in parent groups); 10 weeks (*child:* 10 sessions, 75mins weekly; *parents:* 10 sessions, 90mins weekly)
Horn (1987)	24	Aged 7-11; Generally average to high average intelligence	US	Severe physical impairments; Intellectual deficits; Psychosis in child or parent(s); child currently on medication for control of ADHD symptoms	Conners's Parent Questionnaire - Hyperactivity index, score >15 (2 SDs above mean)	Outpatient psychology clinic (university-based); Doctoral-level clinical psychology graduates	**1. Child self-control training group alone:** Children taught a "problem-solving plan" with self-instructional steps. Token/reward systems and behaviour management techniques (i.e. time outs). **2. Parent training alone:** Taught principles of behaviour management via social learning theory; Bibliotherapy; Individualised behaviour management project initiated with child. **3. Parent training + child self-control training group**	Groups of 8; 8 weeks (8 sessions; 90mins weekly); Separate child and parent groups

Horn (1990)	60	Aged 7-11; 70% male, 30% female; 87% White, 10% Black, 1.5% Hispanic, 1.5% Asian; 52% co-morbid CD; 19% co-morbid ODD	US	Severe physical impairments; Intellectual deficits; Psychosis in either the child or parent(s); Child currently taking medication for ADHD; Evidence of psychopathology in control subjects	DSM-III-R criteria - Clinical interview with parents, CPRS (Score > 2 SDs from mean for Hyperkinesis), CTRS (Score > 2 SDs from mean for Hyperkinesis) and a battery of psychometric tests.	Outpatient psychology clinic (university-based); Clinical psychology training program graduates	**1. Child self-control training group (+ school consultation)**: Similar to *Horn et al. (1987)* with some additions. Children learnt to locate tension within their body and apply relaxation skills. Role plays for problem-solving specified academic and interpersonal scenarios. School meetings: 3 sessions with child's teacher to reinforce skills learnt. **2. Parent training alone (+ school consultation)**: Additions: 3 sessions with teacher at school (start/ middle/end of study) to initiate daily home report card. **3. Parent training + child self-control group (+ school consultation** *(see above)* **4. Controls** (non-ADHD, no intervention)	Groups of 7 (slightly more in parent groups); 12 weeks (12 sessions, 90mins weekly); Separate child and parent groups

Notes: The structure of the table follows the levels of approaches (blue = child-only interventions, pink = dual, orange = multicomponent, green = direct comparison studies); ADHD-I = Inattentive type, ADHD-H/I = Hyperactive/Impulsive type, ADHD-Comb = Combined type; ODD = Oppositional Defiant Disorder; CD = Conduct Disorder; OT = Occupational Therapist; CPRS = Conners Parent Rating Scale; CTRS = Conners Teacher Rating Scale

* Group details refer primarily to child groups (within all levels)

The selected studies included a variety of child-focused group approaches. Some of these (*n* = 7; 32%) were delivered independently, without any other interventions ("child-only groups"). However, some studies (*n* = 4; 18%) evaluated treatments that also involved one other parallel intervention, such as parent or teacher training ("dual interventions"). Many (*n* = 9; 41%) looked at multicomponent approaches, in which three or more parallel interventions (including child-focused groups) were evaluated as a treatment package. Lastly, two studies (9%) directly compared some of these different levels of approaches ("direct comparisons").

General findings

Sample demographics

The demographic data of participants recruited across the selected studies are summarised in Table 3.

Of additional note, nearly all studies reported a minimum of average range intelligence in all participants, or specified IQ < 75-80 as an exclusion criterion. Nearly all studies reported a wide range of additional exclusion criteria, often including history of medical, psychiatric or neurological problems. Studies used various approaches in establishing ADHD diagnosis before participation, all of which were deemed acceptable. These included validated and reliable questionnaires, rating scales, diagnostic interviews (usually combining parent and teacher views) and batteries of psychometric tests. Some studies accepted previous clinical diagnoses from paediatricians or other professionals.

Table 3: Summary of sample demographics across selected studies

Sample size (*n*=22)*	6 - 128
Age range (*n*=22)	
13 or under	19 / 22 studies (86%)
14 or over**	3 / 22 studies (14%)
Gender (*n*=20)	

Males	38 - 100% *(typically over 67%)*
Females***	0 - 62% *(typically under 33%)*
ADHD subtypes (*n*=10)	
ADHD-Comb	50 - 100% *(typically 50 - 62%)*
ADHD-I	31 - 100% *(typically 31 - 41%)*
ADHD-H/I	6 - 100% *(typically 6 - 14%)*
Co-morbid disorders (*n*=13)[a]	
Oppositional Defiant Disorder	14 - 49%
Conduct Disorder	4 - 52%
Anxiety disorders	12 - 28%
Dyslexia	17%
Depression	1 - 6%
Ethnic backgrounds (*n*=12)[b]	
White or Caucasian	35 - 100%
Mixed race	9 - 26%
Hispanic/Latino	1.5 - 24%
Asian/Asian American/Pacific Islander	1.5 - 19%
African-American	6 - 17%
Black	4 - 10%
American Indian	2 - 3%
Caribbean	2%

Note: 'n' refers to the number of studies (out of 22) providing specified information
** Sample sizes were generally small (occasionally medium). Larger studies tended to allocate participants to more treatment conditions*
*** Studies included, but were not limited to, children aged 14 years or older (range: 11 years – adult)*
**** With the exception of one all-female group (Looyeh, Kamali & Shafieian, 2012)*
[a] Zylowska and colleagues' (2008) mixed adult and adolescent sample also reported lifetime co-morbidities of 78% for mood disorders and 34% for anxiety disorders
[b] Exception: So, Leung and Hung (2008) reported an all-Chinese sample

Group treatments

The characteristics of the child-focused group treatments across selected studies are summarised in Table 4.

Of additional note, groups were delivered by various professionals. In clinic settings groups were often run by professionals from a psychology background (undergraduate, masters, doctoral and qualified psychologists; clinical / counselling

psychologists, mindfulness trainers, CBT therapists) but also included researchers and social workers experienced in child therapy. In school settings, groups were again primarily delivered by professionals from a psychology background (undergraduate, doctoral and post-doctoral level; educational psychologists, therapists), but also included Learning Support Professionals (masters-level mental health clinicians) and researchers. Lastly, groups in laboratory classroom settings were run by para-professionals (e.g. nurses, teachers, occupational therapists) and a clinical psychologist.

In terms of the different levels of approaches, parent groups typically received a similar frequency of input and group duration. Group sizes were often larger than child groups. All parent and child groups were run separately (usually including joint elements), with the exception of So, Leung and Hung (2008) in which parents eventually became co-therapists. Teacher training or school consultation tended to be less intensive, although in terms of interactions with the child in the classroom, the potential impact of these interventions would have been daily.

Table 4: Summary of (child) group treatment characteristics across selected studies

Geographical location (*n*=22)	
US	14 / 22 (63%)
The Netherlands	3 / 22 (13%)
Spain	2 / 22 (9%)
Iran	1 / 22 (5%)
Turkey	1 / 22 (5%)
Hong Kong	1 / 22 (5%)
Group setting (*n*=21)	
Outpatient clinic (research or clinical)	12 / 21 (57%)
School	7 / 21 (33%)
Laboratory classroom	2 / 21 (10%)
Child group size (*n*=20)	3 to 10
Child group session length (*n*=22)	30 to 150 minutes
Child group duration (*n*=22)	3 to 14 weeks *(exception: one study ran for 26 weeks/6 months)*
Total child group sessions (*n*=21)	5 to 32
Child group frequency (*n*=18)	
4 x weekly	1 / 18 (6%)

Weekly	11 / 18 (61%)
Fortnightly	6 / 18 (33%)
Use of token/reward systems (*n*=22)	14 / 22 (63%)
Use of response cost procedures (*n*=22)	3 / 22 (13%)

Note: 'n' refers to number of studies (out of 22) providing specified information

Specific group descriptions

Below are descriptions of the specific child groups that were described within each study, which are summarised in Table 5. Full details of interventions reported in each study, including dual and multicomponent treatment packages, are provided in Table 2. All groups were based primarily on behavioural or cognitive-behavioural principles, with one study using a systemic approach (Looyeh, Kamali & Shafieian, 2012). Most studies used techniques such as modelling, guided practice and role play to facilitate the learning process. Methodological quality, study design and outcomes will be discussed in further detail later.

Child-only groups

- **Self-control / cognitive self-regulation and problem-solving training using self-instructional techniques** (Barkley, Copeland & Sivage, 1980; Hinshaw, Henker & Whalen, 1984, Study 1 only).
    - Miranda and Jesús Presentación (2000) used a similar approach but adapted the final 7 sessions in one treatment condition to focus on anger management skills, with an interest in children with and without aggressiveness.
    - Ozcan et al. (2013) ran interpersonal cognitive problem-solving groups (with an emphasis on applying problem-solving techniques within social situations)
- **Stress management** (Gonzalez & Sellers, 2002)

- **Mindfulness meditation training** (Zylowska et al., 2008)
- **Narrative therapy** (Looyeh, Kamali & Shafieian, 2012)

The second study reported in Hinshaw and colleagues (1984) was excluded due to being a summer treatment program.

Dual interventions

All dual approaches involved child and parent groups. Child groups were based on:

- **Psycho-education** (Fields & Hale, 2011)
- **Supporting Teens' Academic Needs Daily-Group** (STAND-G; Sibley et al., 2014): Psycho-education and skills training (academic, organisational, communication).
- **Mindfulness training** (Van de Weijer-Bergsma et al., 2012; Van der Oord, Bögels & Peijnenburg, 2012, combining Mindfulness-Based Cognitive Therapy and Mindfulness-Based Stress Reduction approaches)

Multicomponent interventions

Multicomponent approaches included parent, teacher/school, family sessions and medication (*see Table 2*). Child groups were based on:

- **Problem-solving training** (Bloomquist, August & Ostrander, 1991)
    - With deep muscle relaxation exercises (Horn et al., 1991; Ialongo et al., 1993)
    - With self-instruction techniques, anger management and social skills training (Miranda et al., 2011)
    - With self-monitoring and anger management (So, Leung and Hung, 2008)

    - With relaxation exercises, academic and social skills training (Van der Oord et al., 2007)

- **Child Life and Attention Skills (CLAS) Program** (Pfiffner et al., 2007): Skills training (e.g. independence, problem-solving, social competence) specifically for children with ADHD-I. Pfiffner et al. (2011, 2013) provided a similar program (CLS) that was adapted to suit a school environment.

Direct comparison studies (single vs dual vs multicomponent)

Direct comparison approaches included single versus dual, and dual versus multicomponent approaches. Child groups were based on:

- **Self-control and problem-solving training** (Horn et al., 1987). Horn and colleagues (1990) led a very similar group program, also integrating deep muscle relaxation into the training.

Table 5: Summary of group content

	Description of Groups
Child-Only Groups	**1. Self-control / cognitive self-regulation and problem-solving training using self-instructional techniques** (Barkley, Copeland & Sivage, 1980; Hinshaw, Henker & Whalen, 1984, Study 1 only) ➡ With or without anger management training (Miranda and Jesús Presentación, 2000) ➡ Emphasis on interpersonal contexts (Ozcan et al., 2013) **2. Stress management** (Gonzalez & Sellers, 2002) **3. Mindfulness meditation training** (Zylowska et al., 2008) **4. Narrative therapy** (Looyeh, Kamali & Shafieian, 2012)
Dual Interventions	**1. Psycho-education** (Fields & Hale, 2011) **2. Supporting Teens' Academic Needs Daily-Group** (STAND-G; Sibley et al., 2014): Psycho-education and skills training (academic, organisational, communication) **3. Mindfulness training** (Van de Weijer-Bergsma et al., 2012; Van der Oord, Bögels & Peijnenburg, 2012)
Multicomponent Interventions	**1. Problem-solving training** (Bloomquist, August & Ostrander, 1991) ➡ With deep muscle relaxation exercises (Horn et al., 1991; Ialongo et al., 1993) ➡ With self-instruction techniques, anger management and social skills training (Miranda et al., 2011) ➡ With self-monitoring and anger management (So, Leung and Hung, 2008) ➡ With relaxation exercises, academic and social skills training (Van der Oord et al., 2007) **2. Child Life and Attention Skills (CLAS) Program** (Pfiffner et al., 2007): Skills training (e.g. independence, problem-solving, social competence) specifically for children with ADHD-I. ➡ CLAS Program adapted for school contexts (CLS: Pfiffner et al., 2011, 2013)
Direct Comparison Studies	**1. Self-control and problem-solving training** (Horn et al., 1987) ➡ With deep muscle relaxation exercises (Horn et al., 1990)

Objective 2a: Methodological quality

The EPHPP quality assessment tool was used to evaluate selected studies based on eight categories, six of which were combined to allocate global quality ratings. Table 6 presents the findings of this assessment, which are also summarised below along with descriptions of additional scoring procedures.

Three studies were excluded from quality assessment and evaluation of effectiveness. Barkley, Copeland and Sivage (1980) did not include a pure child group as children also engaged in individual work during the group sessions. Gonzalez and Sellers (2002) recruited part of their sample from a summer camp for children with ADHD. It was not possible to clarify whether these children continued to

attend the summer camp during the study. Zylowska and colleagues (2008) did not analyse results separately for adolescents and adults. Thus, a total of nineteen studies were assessed for methodological quality.

General findings

Overall, a large proportion of studies (9/19) received a Weak global rating. Five were rated as Moderate and five as Strong. In terms of the levels of approaches, all child-only groups (4/4) scored as Weak. Dual interventions scored as Weak (3/4) and Strong (1/4). Multicomponent interventions were rated as Strong (3/9), Moderate (4/9) and Weak (2/9). Lastly, direct comparison studies were scored as Strong (1/2) and Moderate (1/2).

Selection bias

In terms of scoring, community/non-referred samples were seen as most likely to encompass children with a range of ADHD severities and presentations, thus being rated as “very likely” to be representative. Clinical referrals for children in need of treatment to a medical or psychological clinic or research facility were scored as “somewhat likely” to be representative. Very specific pools of subjects (e.g. university centre mailing lists) were scored as “not likely” to be representative. Studies which recruited subjects from several different methods (as described above) were rated as “very likely” to be representative.

Most studies (*n* = 13) scored as Moderate for selection bias, with five scoring as Strong and one as weak (Sibley et al., 2014).

Study design

Given that most studies were classified as Randomised or Controlled Clinical Trials, many (*n* = 15) received ratings of Strong. These studies included active, waitlist/no

treatment or treatment as usual control groups (*for details, see Table 2*). The remaining cohort studies were rated as Moderate.

Confounders

Nine studies were rated as Strong and unlikely to be influenced by confounders. Nine were rated as Weak (mostly child-only and dual interventions) and one as Moderate.

Blinding

In terms of scoring, studies reported various types of blinding (e.g. blind to time point, treatment condition, sample characteristics, research question). Scoring was completed on the basis of the majority, so for example if outcome raters were blind to time point and treatment, but not sample characteristics, they were scored as blinded overall. If the majority was unclear, then unclear was scored. If types of blinding were at least half unclear, unclear was recorded (unless outcome measures were weighted much heavier for one type of rater). If studies described equal amounts such as one third blinding, one third not blinded and one third unclear, unclear was scored. If studies were half blinded and half not blinded, they received a score of Moderate. All outcome raters (e.g. parents, teachers, child self-report) were considered when allocating blinding scores, rather than just examiners or researchers alone, as these were all seen as potentially being vulnerable to expectancy biases and thus influencing treatment outcomes.

No studies reported whether participants were aware of the research question, hence there were no Strong ratings in this category. Most (*n* = 11) were scored as Moderate, and eight as Weak.

Data collection methods

Most studies (*n* = 15) used dependent measures that were known or reported to be valid and reliable. For others this was not clear or not reported, with three being rated

as Weak and one as Moderate. Some exceptions were made if measures for which validity and/or reliability was unclear were not considered relevant for the present review (e.g. parent measures not at all related to possible impact of child groups).

Withdrawals and drop-outs

These ratings were given conservatively, scoring according to the lowest rate of participants completing the study reported, whether at post-test or follow-up. Most studies (*n* = 12) were assessed as Strong. The remaining studies were scored as Moderate (*n* = 4) and Weak (*n* = 3), the latter of which was given when studies failed to report withdrawals or drop-outs at all.

Intervention integrity

This category did not contribute to global ratings, thus scores were not given. However, the integrity of treatments (e.g. no contamination from other influences, whether delivery was standardised and so on), was still of interest descriptively. All except three of the nineteen studies were considered to have good intervention integrity. In Fields and Hale (2011), contamination was likely as the intervention was an adjunct to treatment as usual. In Pfiffner et al. (2007, 2011) contamination was unlikely, however interventions were not consistent as changes were made to the program with different cohorts (e.g. number of sessions varied from 8 to 10).

Analysis

This category did not contribute to global ratings, thus scores were not given. Nearly all studies (17/19) applied appropriate statistical methods for the study design. Sibley et al. (2014) described effect sizes for some quantitative data, but did not engage in any other relevant statistical analyses. Van de Weijer-Bergsma et al. (2012) used paired *t*-tests when a repeated-measures ANOVA may have been more appropriate to examine participant progression from pre-test through to follow-up.

Seven studies reported applying either an intention-to-treat analysis or analysis of 100% of participant outcome data. Eight studies did not report using an intention-to-treat analysis and did not analyse all participant data. Four studies did not specify this information.

Table 6: Assessment of methodological quality (n = 19)

Primary author (date)	Selection bias	Study design	Confounders	Blinding	Data collection methods	Withdrawals & drop-outs	Intervention Integrity*	Analyses*	Global rating
Hinshaw (1984) - Study 1 only	*Moderate (2)* - Recruited from local schools, paediatricians and newspaper adverts; Unclear % of selected individuals agreed to participate	*Strong (1)* - Randomised Controlled Trial	*Weak (3)* - Control of confounders was not described	*Moderate (2)* - Children and families unaware if placebo or medication; Observers blind to medication status and to time point, but not to groups; Unclear if study participants were aware of the research question	*Weak (3)* - Reliability and validity of outcome measures (observational coding systems) were not described	*Strong (1)* - Post-intervention data was collected for all cases	All participants received the planned interventions. Good intervention consistency: All group sessions were carefully scripted and standardized. Contamination unlikely.	Appropriate statistical methods for study design; Not intention-to-treat analysis	***Weak (3)***
Looyeh (2012)	*Moderate (2)* - Children referred by schools to clinic; Unclear % of selected individuals who agreed to participate	*Strong (1)* - Controlled clinical trial	*Weak (3)* - Control of confounders was not described	*Moderate (2)* - Single-blind: Teachers not aware treatment was occurring (they were the only raters on outcome measures); Unclear if participants were aware of the research question.	*Strong (1)* - Primary outcome measure valid and reliable	*Weak (3)* - Not described	All participants received the planned interventions. Good intervention consistency: Clear plans for every group session. Contamination unlikely.	Appropriate statistical methods for study design; Unclear if intention-to-treat analysis was carried out.	***Weak (3)***
Miranda (2000)	*Moderate (2)* - Children selected by school psychologists at 15 schools; Unclear % of selected individuals who agreed to participate	*Strong (1)* - Controlled clinical trial	*Weak (3)* - Control of confounders was not described	*Moderate (2)* - Therapists running groups were not told which children were labelled aggressive or nonaggressive; Parents aware of treatment condition. Unclear if teachers blind. Unclear if participants were aware of the research question.	*Weak (3)* - Most outcome measures valid and reliable. Unclear for one parent-rated and one teacher-rated measure.	*Weak (3)* - Not described	All participants received the planned interventions. Good intervention consistency: Structured programmes followed, carried out by the same expert therapists. Contamination unlikely.	Appropriate statistical methods for study design; Unclear if intention-to-treat analysis was carried out.	***Weak (3)***
Ozcan (2013)	*Moderate (2)* - Children recruited from two schools (recruitment process unclear); 100% of selected individuals agreed to participate	*Moderate (2)* - Cohort design (pre-post)	*Weak (3)* - Control of confounders was not described	*Weak (3)* - Outcome assessors (parents and teachers) were aware of the intervention. Unclear if study participants were aware of the research question.	*Strong (1)* - Outcome measures valid and reliable	*Strong (1)* - Post-intervention data was collected for all cases	All participants received the planned interventions. Good intervention consistency: Structured program and training. Contamination unlikely.	Appropriate statistical methods for study design; All participants included in analysis (100% completers)	***Weak (3)***

Fields (2011)	*Moderate (2)* - Children recruited through an outpatient family medicine centre; Unclear % of selected individuals who agreed to participate	*Strong (1)* - Controlled clinical trial	*Strong (1)* - No significant pre-test group differences on demographic data. Some significant differences for treatment history, but addressed in analyses	*Moderate (2)* - Parents aware of intervention status (unclear if teachers were aware); Unclear if study participants were aware of the research question	*Strong (1)* - Primary outcome measure valid and reliable	*Strong (1)* - Post-intervention data was collected for all cases	All participants received the planned interventions. Those with inadequate attendance were excluded from analyses. Fairly good intervention consistency (rough session plans but unclear if fully manualised/adhered to). Contamination likely: Intervention was adjunct to treatment as usual	Appropriate statistical methods for study design; Intention-to-treat not described, but all participants included in analysis (100% completers)	***Strong (1)*** 
Sibley (2014)	*Weak (3)* - Children recruited from the mailing list of a large university research centre; 100% of selected individuals agreed to participate	*Strong (1)* - Controlled clinical trial	*Weak (3)* - Control of confounders was not described	*Moderate (2)* - Most outcome measures objective and academic. Parents and children completed improvement rating scales - Both aware of treatment condition. Unclear if aware of research question.	*Moderate (2)* - Not applicable for objective academic outcomes. One self- and parent-rated improvement scale had some support for validity, but reliability was unclear.	*Strong (1)* - 89% of participants completed the study (3/28 drop-outs)	All participants received the planned intervention. Good intervention consistency (manualized treatments, fidelity checks). Good attendance on average. Contamination unlikely.	Poor statistical analysis of quantitative data; Not intention-to-treat, completers only (89%)	***Weak (3)*** 
Van der Oord (2012)	*Moderate (2)* - Children referred to outpatient clinics by school psychologists, paediatricians or GPs; 92% of selected children who agreed to participate	*Strong (1)* - Controlled clinical trial	*Weak (3)* - Control of confounders was not described	*Weak (3)* - Parents not blind (received intervention and rated outcomes); Unclear if teachers were blind. Unclear if study participants were aware of the research question.	*Strong (1)* - Outcome measures valid and reliable.	*Strong (1)* - 82% of participants completed the study (4/22 drop-outs)	All participants received the planned intervention. Good intervention consistency (manualized treatments, supervision, training). Good attendance rates. Contamination unlikely.	Appropriate statistical methods for study design; Intention-to-treat analysis was carried out.	***Weak (3)*** 
Van de Weijer-Bergsma (2012)	*Moderate (2)* - Children referred to an academic centre; Unclear % of selected children who agreed to participate	*Moderate (2)* - Cohort design (pre-post)	*Weak (3)* - Medication controlled for in method and analysis; Control of other possible confounders not described	*Weak (3)* - Parents and children received the intervention and completed outcome measures. Unclear what teachers were informed. Unclear if participants were aware of the research question.	*Strong (1)* - Outcome measures valid and reliable.	*Moderate (2)* - 100% of participants completed post-test measures; 60% completed follow-up measures (4/10 drop-outs)	All participants received the planned intervention. Good intervention consistency (manualized treatments, training, supervision). Contamination unlikely.	Adequate but not ideal statistical methods for study design; Intention-to-treat analysis was not carried out	***Weak (3)*** 

Bloomquist (1991)	*Strong (1)* - Wide-scale screening in schools, non-referred; 90% of selected individuals agreed to participate	*Strong (1)* - Controlled clinical trial	*Strong (1)* - No indication of any confounders (no significant pre-intervention group differences)	*Moderate (2)* - Outcome assessors for observations were blinded; Unclear if participants were aware of the research question	*Weak (3)* - One primary observational measure showed some reliability, but validity of coding scales not discussed; Others reliable and valid	*Strong (1)* - Post-intervention data was collected for all cases	All participants received planned interventions. Parents with inadequate attendance excluded from analyses. Intervention consistency good (manualised, fidelity monitored). Contamination unlikely.	Appropriate statistical methods for study design; Not intention-to-treat (only 70% of cases included in analyses)	***Moderate (2)***
Horn (1991)	*Moderate (2)* - Referred to university-based clinic; Controls recruited through newspaper adverts and school newsletters; 91% of selected individuals agreed to participate	*Strong (1)* - Controlled clinical trial	*Strong (1)* - Some significant pre-intervention group differences, but analyses showed no significant impact on treatment response.	*Moderate (2)* - Child and parents blind to medication but not group intervention. Teachers most likely blind to medication status but not group treatment. Unclear if examiners were blind. Unclear if participants were aware of the research question.	*Strong (1)* - Nearly all outcome measures valid and reliable. Unclear for one measure, although not important for the key findings of the study.	*Strong (1)* - 85% of participants completed the study (18/117 drop-outs)	All participants received the planned interventions. Good intervention consistency: Thorough training, weekly supervision and manualised treatments for both groups. Contamination unlikely.	Appropriate statistical methods for study design; Intention-to-treat analysis was carried out.	***Strong (1)***
Ialongo (1993) - *9 month follow up of Horn (1991)*	*Moderate (2)* - Referred to university-based clinic; Controls recruited through newspaper adverts and school newsletters; 91% of selected individuals agreed to participate	*Strong (1)* - Controlled clinical trial	*Strong (1)* - Some significant pre-intervention group differences, but analysis showed no significant impact on treatment response.	*Moderate (2)* - Child and parents blind to medication but not group intervention. Teachers most likely blind to medication status but not group treatment. Unclear if examiners were blind. Unclear if participants were aware of the research question.	*Strong (1)* - Nearly all outcome measures valid and reliable. Unclear for one measure, although not important for the key findings of the study.	*Moderate (2)* - 79% of participants completed the study (25/117 total drop outs from pre-test to 9-month follow-up)	All participants received the planned interventions. Good intervention consistency: Thorough training, weekly supervision and manualised treatments for both groups. Contamination unlikely.	Appropriate statistical methods for study design; Intention-to-treat analysis was carried out.	***Strong (1)***
Miranda (2011)	*Strong (1)* - Referred by psychopedagogy services and parent associations in 24 schools; 100% of selected children agreed to participate	*Strong (1)* - Randomised Controlled Trial	*Strong (1)* - Several significant pre-test group differences controlled for in analyses. Two variables not further analysed.	*Moderate (2)* - Blinding not described. Unclear if study participants were aware of the research question.	*Strong (1)* - Outcome measures valid and reliable	*Weak (3)* - Not described	All participants received the planned interventions. Good intervention consistency: Well-structured programs for all treatment components. Good attendance. Contamination unlikely.	Appropriate statistical methods for study design; Unclear if intention-to-treat analysis was carried out.	***Moderate (2)***

Pfiffner (2007)	*Strong (1)* - Recruited from schools via presentations / mailings to school personnel; ADHD outpatient clinic and parent recommendations; 100% of selected individuals agreed to participate	*Strong (1)* - Controlled clinical trial	*Strong (1)* - No indication of any confounders (no significant pre- group differences, or differences between waitlist and treatment as usual control groups)	*Weak (3)* - Outcome assessors (parents and teachers) were aware of the intervention. Unclear if study participants were aware of the research question.	*Strong (1)* - Outcome measures valid and reliable	*Moderate (2)* - 96% of participants completed pre to post-test measures (3/69 drop-outs); 78% of participants completed pre to follow-up measures (15/69 drop-outs)	All participants received the planned intervention. Partially good intervention consistency (manualized treatments, fidelity checklists), however some changes made to program with different cohorts (e.g. number of sessions). Good attendance. Contamination unlikely.	Appropriate statistical methods for study design; Intention-to-treat analysis was carried out.	***Moderate (2)*** 
Pfiffner (2011)	*Strong (1)* - ADHD children identified by Learning Support Professionals in five participating schools; 95% of selected individuals agreed to participate	*Moderate (2)* - Cohort design (pre-post)	*Weak (3)* - Control of confounders was not described	*Weak (3)* - Outcome assessors (parents and teachers) were aware of the intervention. Unclear if study participants were aware of the research question.	*Strong (1)* - Outcome measures valid and reliable	*Strong (1)* - 95% of participants completed the study (2/37 drop outs)	All participants received the planned intervention. Partially good intervention consistency (manualized treatments), however changes made to program with different cohorts. Contamination unlikely.	Appropriate statistical methods for study design; Unclear if intention-to-treat analysis was carried out.	***Weak (3)*** 
Pfiffner (2013)	*Strong (1)* - ADHD children identified by Learning Support Professionals in nine participating schools; 100% of selected individuals agreed to participate	*Moderate (2)* - Cohort design (pre-post)	*Weak (3)* - Control of confounders was not described	*Weak (3)* - Outcome assessors (parents and teachers) not blind. Observational data: Researchers blind to group assignment and purpose of the study. Unclear if study participants were aware of the research question.	*Strong (1)* - Outcome measures valid and reliable.	*Strong (1)* - 95% of participants completed the study (3/60 drop-outs)	All participants received the planned intervention. Good intervention consistency (training, supervision, manualized treatments and fidelity checks). Good attendance on average. Contamination unlikely.	Appropriate statistical methods for study design; Not intention-to-treat, completers only (95%)	***Weak (3)*** 
So (2008)	*Moderate (2)* - Children referred to a community child psychiatric clinic; 100% of selected individuals agreed to participate	*Strong (1)* - Randomised Controlled Trial	*Moderate (2)* - No significant pre-test group differences on demographic data. Medication adherence significantly better in combined group (not controlled for in analysis).	*Moderate (2)* - Pre-intervention assessments done by researchers blind to study hypotheses and treatments. However, for main outcome measure, parents not blind and unclear if teachers were blind. Unclear if study participants were aware of the research question.	*Strong (1)* - Outcome measures valid and reliable.	*Strong (1)* - 86% of participants completed post-test measures (13/90 drop-outs). 84% of participants completed follow-up (14/90 drop-outs pre to follow-up)	All participants received the planned intervention. Good intervention consistency (manualized treatments, training, supervision with audiotapes, fidelity checks). Contamination unlikely.	Appropriate statistical methods for study design; Intention-to-treat analysis was carried out.	***Strong (1)*** 

Van der Oord (2007)	*Moderate (2)* - Children referred to outpatient clinic by school psychologists/ paediatricians/GPs; Unclear % of selected children who agreed to participate	*Strong (1)* - Controlled clinical trial	*Strong (1)* - No indication of any confounders (no significant pre-intervention group differences)	*Weak (3)* - Parents and teachers not blind (took part in interventions and rated outcomes). Unclear whether examiners for child outcome measures were blind. Unclear if study participants were aware of the research question.	*Strong (1)* - Outcome measures valid and reliable.	*Strong (1)* - 94% of participants completed the study (3/50 drop-outs)	All participants received the planned intervention. Good intervention consistency (manualized treatments, fidelity checklist, supervision, training). Medication compliance checks. Contamination unlikely.	Appropriate statistical methods for study design; Not intention-to-treat, completers only (94%)	***Moderate (2)***
Horn (1987)	*Moderate (2)* - Referred to university-based clinic by paediatricians, schools & parents themselves; 100% of selected individuals agreed to participate	*Strong (1)* - Controlled clinical trial	*Strong (1)* - No indication of any confounders (no significant pre-intervention group differences)	*Moderate (2)* - Observers blind to time point and treatment group; Teachers blind to treatment condition, but parents were not. Unclear if study participants were aware of the research question.	*Strong (1)* - Nearly all measures valid and reliable. Unclear for one parent outcome measure, however this scale was not of interest in the present review.	*Moderate (2)* - 79% of participants completed the study (5/24 drop-outs)	All participants received the planned interventions. Good intervention consistency: Training, weekly supervision and manualized treatments for both groups. Contamination unlikely.	Appropriate statistical methods for study design; Not intention-to-treat, completers only (79%)	***Strong (1)***
Horn (1990)	*Moderate (2)* - Referred to university-based clinic; Controls recruited through newspaper adverts and school newsletters; 100% of selected individuals agreed to participate	*Strong (1)* - Controlled clinical trial	*Strong (1)* - No indication of any confounders (no significant pre-intervention group differences or covariates)	*Weak (3)* - Examiners blind to treatment status/ condition and time point. Parents and teachers not blind. Unclear if study participants were aware of the research question.	*Strong (1)* - Nearly all outcome measures known to be valid and reliable. Unclear for one observational coding measure. However, this was not of interest in the present review.	*Strong (1)* - 87% of participants completed the study (8/60 drop-outs)	All participants received the planned interventions. Good intervention consistency: Thorough training, weekly supervision (with audiotapes of sessions) and manualized treatments for both groups. Contamination unlikely.	Appropriate statistical methods for study design; Not intention-to-treat, completers only (87%)	***Moderate (2)***

Note: Data is structured by level of approach, with clear separations (from top to bottom: child-only, dual, multicomponent and direct comparison studies)
* Intervention Integrity and Analyses sections are not included in global ratings but are provided for descriptive purposes

Objectives 2b and 2c: Evaluation of effectiveness and support for groups

It was decided that only studies with Moderate – Strong methodological quality would be included in the evaluation of effectiveness. Nine studies with Weak methodology were therefore excluded (Hinshaw, Henker & Whalen, 1984; Looyeh, Kamali & Shafieian, 2012; Miranda & Jesús Presentación, 2000; Ozcan et al., 2013; Sibley et al., 2014; Van der Oord, Bögels & Peijnenburg, 2012; Van de Weijer-Bergsma et al., 2012; Pfiffner et al., 2011 & 2013). Additional data extracted from the ten studies progressing to the evaluation of effectiveness included: Time points of measurement, medication status, outcome measures, main findings, effect sizes (when reported), main limitations and an overall indication of support for the child groups[1]. This information is displayed in Table 7 and detailed below. This information for the nine excluded studies can be found in Appendix 4.

Most studies had small sample sizes and may have been under-powered to detect treatment effects (particularly those with small-medium effect sizes).

[1] Short-term effects were based on pre- to post-test findings; Long-term effects were based on follow-up findings of any length.

Table 7: Evaluation of effectiveness (n = 10)

Primary author (date)	Time points	Intervention summary	Medication status	Outcome measures	Main findings	Reported effect sizes*	Main limitations	Support for groups?**
Fields (2011)	Pre, post (4 weeks after groups)	1. Child and parent **psycho-education groups** 2. Waitlist control	More children in experimental group medicated than controls (analysis indicated no significant impact on outcomes)	*NICHQ Vanderbilt Assessment Scale* - Brief rating scale for parents and teachers (ADHD symptoms and school performance scales used)	Children in the treatment and control groups both improved significantly over time from the parent (not teacher) perspective, in terms of ADHD symptoms and school performance. Significant main effect of treatment group found for parent-rated school performance only, with those in the treatment group making modest gains compared with controls.		Only 1 outcome measure; Parent ratings could be due to expectancy effects; Treatment contamination (adjunct to treatment as usual); Small *n*	Limited support (short-term)
Bloomquist (1991)	Pre, post, 6 weeks	1. **Multicomponent Cognitive-Behavioural Therapy (MCBT)** 2. Teacher only intervention 3. Waitlist control	Any children on psychostimulant medication were excluded	*Child:* 1. On/off-task behaviour, coded classroom observations 2. PHSC *Teachers:* 1. Self-Control Rating Scale 2. CTRS 3. Walker-McConnell Scale of Social Competence and School Adjustment	MCBT condition significantly better than the other conditions at improving observed off-task/disruptive behaviour. Significance not maintained at follow-up. There were no other significant group x time differences on other measures.		Validity of observational coding scales for main finding unclear; Small *n* for analysis (30% of cases invalid)	Some support for short-term effects
Horn (1991)	Pre, post	1. Medication placebo 2/3. Low/High dose medication 4. Medication placebo + behavioural parent training + **child self-control group** + school consultation 5/6. Low/High dose + behavioural parent training + child self-control group + school consultation 7. Non-ADHD controls	Children not on medication prior to study, or if they were a 2-week discontinuation period was required; Medication manipulated experimentally	*Parent/teacher:* 1. CBCL 2. SNAP 3. CPRS 4. CTRS 5. Teacher checklists of Children's Peer Relations and Social Skills 6. Personality Inventory for Children-Revised, Family Relations subscale (parents) *Child:* 1. CPT (attention/ impulsivity) 2. Clinic-based observations (motor activity / inattention in 15min academic setting) based on the SOAPS 3. WRAT-R 4. PHSC scales 5. NSLCS	No support for the superiority of combined behavioural treatments (BTs) relative to medication alone. *Parent-rated measures:* Most treatment groups (including placebo only) improved significantly at post-test. *Teacher measures*: Some limited evidence that effects of high dose medication on ADHD symptoms could be achieved by combining low dose with BTs. *Child measures*: Significant effects of medication over time for academic, impulsivity and self-concept outcomes. No significant effects for BT conditions. *Observational data*: Significant effects of medication over time for off-task behaviour. Some support for medication (and on one occasion combined BT + low dose) normalising hyperactivity, impulsivity/attention and self-concept relative to control subjects.		Expectancy effects (parents/ teachers/ children); No BT- only group (all included placebo or medication)	Little support relative to medication in the short-term (except possibly allowing lower medication dose for same effect)
Ialongo (1993) - *9 month follow up of Horn (1991)*	9m	*See Horn (1991)*	*See Horn (1991)* Any medication was withdrawn immediately after post-test assessments	*See Horn (1991)*	In contrast to the medication alone condition, the combined BTs resulted in significantly greater improvement in follow-up parent ratings of ADHD symptoms and externalising behaviours. However, this was not supported by child, teacher or observational follow-up measures. Overall trend with these latter measures towards an erosion of treatment gains seen from pre- to post-test, across all treatment conditions.		Expectancy effects (parents)	Very little support for long-term benefits

Miranda (2011)	Pre, post	1. **Cognitive-behavioural child groups + parent group** + teacher training 2. No treatment control group	No participants were prescribed medication for ADHD for the duration of the study	*Executive functioning (child measures):* 1. CPT 2. Stroop test (adapted) 3. Working Memory Sentences 4. WISC-R (Inverse Digits only) 5. Temporal Spatial Recall Task 6. Tower of London 7. Wisconsin Card Sorting Test *ADHD qurs:* Parents / teachers (DSM-IV-TR criteria)	Children in the treatment group improved significantly on almost all executive functioning variables (1. CPT attention 2. Stroop interference control 3. Verbal and 4. Visuospatial working memory 5. Planning), in relation to the untreated control group. Furthermore, significant improvements were observed in the parents' (6,7) and teachers' (8,9) behavioural ratings of attention and hyperactivity/impulsivity (respectively) in the treated group only.	Med-Large: 1. η^2 = .288 2. η^2 = .198 3. η^2 = .125 4. η^2 = .396 5. η^2 = .135 6. η^2 = .412 7. η^2 = .282 8. η^2 = .329 9. η^2 = .282	Small *n*; No comparison with other active treatments; No follow-up	Good support (short-term) with mostly large effect sizes; Some limitations
Pfiffner (2007)	Pre, post, 3-5m	1. **Child Life and Attention Skills (CLAS) Program** 2. Waitlist control group or Treatment as usual	Only two participants (3%) were taking medication for ADHD when recruited. This was required to remain stable throughout the study.	*Parent/teacher:* 1. CSI (DSM-IV Inattention only) 2. The sluggish cognitive tempo (SCT) Scale 3. Social Skills Rating System 4. COSS 5. Clinical Global Impressions - Improvement	Children randomized to the CLAS Program were reported to have significantly improved inattention (1. symptoms and 2. severity), 3. sluggish cognitive tempo, 4. social and 5. organisational skills, relative to the control group. Gains were maintained at follow-up. On an improvement scale, parents and teachers reported significantly greater improvement at post-treatment for the treated than the control group.	Med-large: 1. η^2p=.184 F-up: η^2p=.09 2. η^2p=.190 F-up: η^2p=.11 3. η^2p=.224 F-up: η^2p=.056 (borderline medium) 4. η^2p=.112 5. η^2p=.173	Expectancy effects; ADHD-Inattentive type only; Small *n*	Good support from outcomes (large effect sizes in short-term and medium in long-term), with limitations
So (2008)	Pre, post, 6m, 12m	1. Methylphenidate 2. Methylphenidate + **Behavioural Treatment** (BT; child groups and parent training)	All participants had never taken stimulant medication for more than 2 weeks; Medication manipulated experimentally	*1. Strengths and Weaknesses of ADHD Symptoms and Normal Behaviours (SWAN) Rating Scale:* Behaviours related to ADHD and ODD, parent and teacher-rated [SWANc composite score = Average of all SWAN items (including Defiance) from parents and teachers]	The combination of BT and low-dose methylphenidate was significantly more effective than methylphenidate-only in reducing ADHD and ODD symptoms at post-treatment. At follow-ups, the benefits of the combined treatment were maintained, while the methylphenidate-only group caught up in improvement in ADHD symptoms.	Medium (combined vs medication at post-test) SWANc: *d*=.62; ADHD symptoms: *d*=.55; ODD symptoms: *d*=.66	Expectancy effects; One main outcome measure; No medication-free groups; Group differences in medication adherence not controlled	Some support (short and long-term), but only as an adjunct to medication (some limitations)
Van der Oord (2007)	Pre, post	1. Methylphenidate 2. Methylphenidate + **multimodal behaviour therapy (child groups** + parent training + teacher training)	All participants had never taken methylphenidate; Medication manipulated experimentally	*Parents (P) / teachers (T):*1. DBDRS (DSM-IV criteria, 4 scales: Inattention, Hyperactivity/ Impulsivity, ODD & CD 2. Social Skills Rating Scale 3. PSI *Child*: 1. Self-Perception Profile for Children, global Self Worth subscale 2. State Trait Anxiety Inventory for Children	Both treatment conditions yielded significant improvements on all outcome domains. No significant differences were found between both treatments. Thus, no evidence for the additive effect of multimodal behaviour therapy next to optimally titrated methylphenidate.	Pre-post (main effect of time, not group): Large for P & T measures: η^2 =.21-.59; Med-large for child measures: η^2 = .09-.25	Small *n*; Expectancy effects; No medication-free treatment or control groups	No support when compared to medication

Horn (1987)	Pre, post, 1m	1. **Child self-control training** 2. Parent training 3. Parent training plus child self-control training	No participants were currently being prescribed medication for ADHD	*Child:* 1. NSLCS 2. Children's Perceived Self-Control Scale 3. PHSC (abbreviated) 4. Kaganys Matching Familiar Figures (attention and impulsivity) 5. 30min classroom observations, 5 areas (e.g. off-task behaviour) 6. WRAT *Parent:* Connors Parent Qur *Teacher:* Conners's Teacher Qur	All three groups improved significantly on numerous child and parent-reported outcomes (e.g. hyperactivity, self-control/concept, impulsivity; not conduct, classroom or academic). No evidence that combining parent and child groups leads to greater treatment effects. Multiple-regression showed that children who were more reflective and had an internal locus of control demonstrated the greatest improvements. *Follow-up:* Gains maintained on parent measures (hyperactivity, total problems, conduct and anxiety). Some gains maintained on child measures (PHSC, matching figures). Significantly greater decrease in hyperactivity scores for self-control instruction alone group.		Small *n*; Expectancy effects (parents); Short follow-up period; No waitlist control or control for therapist contact	Good support (short and long-term), some limitations
Horn (1990)	Pre, post, 8m	1. **Child group self-control training** (+ school consultation): 2. Parent training alone (+ school consultation) 3. Parent training plus child self-control training (+ school consultation) 4. Control group (non-ADHD, no intervention)	No participants were currently being prescribed medication for ADHD	*Parent/teacher:* 1. CBCL 2. CPRS 3. CTRS 4. Teacher Self-Control Rating Scale 5. Personality Inventory for Children-Revised, Family Relations subscale (parent) *Child:* 1. CPT 2. WRAT-R 3. PHSC	Children in all three treatment conditions demonstrated significant improvements at post-test on numerous parent and teacher (not child) measures. Maintained at follow-up in parent measures only. No significant Treatment Group x Time interactions were found. Children receiving the combined treatment improved to the point where, at follow-up, they were no longer significantly different on the PHSC compared to normal controls.		Expectancy effects; Small *n* (for each condition)	Good support (short and long-term), some limitations

Notes: m = month; Qur = Questionnaire; ODD = Oppositional Defiant Disorder; CD = Conduct Disorder. BRIEF = Behaviour Rating Inventory of Executive Function; CBCL = Child Behaviour Checklist; CPRS = Connors Parent Rating Scale; COSS is Children`s Organizational Skills Scale; CPT = Continuous Performance Test; CSI = Child Symptom Inventory; CTRS = Conners Teacher Rating Scale; DBDRS = Disruptive Behaviour Disorders Screening and Rating Scale; NSLCS = Nowicki-Strickland Locus of Control Scale for Children; PHSC = Child Report Piers-Harris Self-Concept Scale; PSI = Parenting Stress Index; SOAPS = Structured Observation of Academic and Play Settings; WISC-R = Wechsler Intelligence Scale for Children, Revised; WRAT-R = Wide Range Achievement Test-Revised

*Effect sizes reported are described according to conventional estimates provided by Cohen (1988): For η^2 (eta-squared) and η^2p (partial eta-squared): 0.01-0.05 = small, 0.06-0.13 = medium, >0.14 = large; For d: <0.4 = small, 0.4–0.8 medium and >0.8 = large

**Short-term effects = pre-post findings; Long-term effects = Follow-up of any length (not including post-intervention)

Child-only groups

All child-only groups were all assessed to have Weak methodology and were thus excluded from this evaluation.

Dual interventions

All studies were assessed to have Weak methodology with the exception of Fields and Hale (2011), which was rated as Strong. One dependent measure was applied at pre- and post-intervention to assess ADHD symptoms and school performance outcomes. Support for the child and parent psycho-educational groups was limited (short-term), as children in both the treatment and control groups improved significantly over time. Children in the treatment group were rated as improving significantly on school performance scales compared to controls, however this was also supported by parent-ratings. Medication status did not have a significant impact on outcomes.

Multicomponent

Two studies (Pfiffner et al., 2011 & 2013) were assessed to have Weak methodology and were thus excluded from this evaluation. All remaining studies had a fairly similar duration of group treatment (10 – 12 weeks), with the exception of So, Leung and Hung (2008) which was six months.

The remaining seven studies provided interventions which, although varied, all followed a cognitive-behavioural framework and were conceptually similar in many ways (e.g. frequent inclusion of problem-solving techniques, particularly within the context of social skills development, self-control and anger management). Four studies manipulated medication experimentally, while the other studies either required that medication was kept stable (Pfiffner et al., 2007) or children were unmedicated (Miranda et al., 2011; Bloomquist, August & Ostrander, 1991). Many studies reported the use of daily home-school report card systems, which involved updates of target

behaviours being given each day by teachers to parents (often involving rewards for progress), encouraging communication and joint working.

Bloomquist, August and Ostrander (1991) utilised child and teacher-rated outcome measures, as well as classroom behavioural observations, to assess ADHD symptoms, self-control, self-concept, social competence and school adjustment. Some limited (short-term) support for multi-component cognitive-behaviour therapy (MCBT; child, parent and teacher interventions) was provided. Children who received MCBT showed significantly improved on/off-task classroom behaviour than those in teacher-only or waitlist control conditions. However, the changes were not maintained at follow-up and there were no other significant findings on other measures. One limitation of the study was that the validity for the main observational coding scale was unclear.

Horn et al. (1991), along with Ialongo et al.'s (1993) 9-month follow-up, utilised a large number of parent, teacher and child-dependent outcome measures, assessing ADHD symptoms, behaviour, interpersonal skills, locus of control, self-concept and academic performance. At post-intervention, there was little support for the superiority of the Psychosocial Intervention Package (PIP; child self-control group, parent training, school consultation) relative to medication alone, at least in the short-term. One exception to this involved teacher-rated ADHD symptoms, in which that the effect of high dose medication could be achieved by combining low dose with PIP. On parent-rated measures, children in all conditions (even placebo) improved. Medication was superior to PIP on child academic, attention, impulsivity and self-concept outcomes. Ialongo et al. (1993) provided some limited support for the long-term benefits of PIP. Children who received PIP improved significantly more on parent-rated ADHD symptoms and externalising behaviours compared to those in medication alone conditions. However, this was not supported by child, teacher or observational measures.

Miranda et al. (2011) were interested in the impact of a multicomponent cognitive-behavioural intervention (MCBI; child, parent, teacher) on children's executive functioning (standardised tests) and ADHD symptoms. MCBI received good support for short-term benefits, with medium to (mostly) large effect sizes. Children in the MCBI group improved significantly on almost all executive functioning variables, as well as parent and teacher-rated ADHD symptoms, in relation to controls.

Pfiffner et al. (2007) evaluated the benefits of the Child Life and Attention Skills (CLAS; child, parent, family, teacher) program in relation to parent and teacher-rated measures of inattention, sluggish cognitive tempo, social/organisational skills and clinical improvement. CLAS received good support, demonstrating large and medium effect sizes in the short and long-term respectively. Children in the CLAS program improved significantly in all areas, relative to the control group. Gains were maintained at follow-up.

So, Leung and Hung (2008) measured parent and teacher-rated ADHD symptoms and ODD behaviours to evaluate the benefits of providing a behavioural treatment program (BT; child, parent, teacher) as an adjunct to medication. BT received limited support in the short and long-term. The combination of BT and low-dose methylphenidate was significantly more effective than methylphenidate-only in reducing ADHD and ODD symptoms at post-treatment, in which medium effect sizes were observed. Benefits were maintained at follow-up, although the medication-only group caught up in improvement in ADHD symptoms. One limitation was that medication adherence was significantly better in the combined group, which was not controlled for in statistical analyses.

Van der Oord et al. (2007) utilised several parent and teacher-rated measures of ADHD symptoms, behaviour (ODD / CD), social skills and parenting stress, as well as child-rated measures of self-worth and anxiety. The evaluations were used from pre to post-treatment to assess the additive effect of multimodal behaviour therapy (MBT;

child, parent, teacher) to medication, compared with medication alone. There was no evidence for the additive benefit of MBT compared with medication alone. Children in both conditions improved significantly in all domains, with no significant differences between treatments.

Direct comparisons

These studies were assessed to have Strong (Horn et al., 1987) and Moderate (Horn et al., 1990) methodological quality. The earlier study by Horn and colleagues had a shorter group duration than the latter (8 vs 12 weeks), however both studies were conducted weekly for 90 minutes.

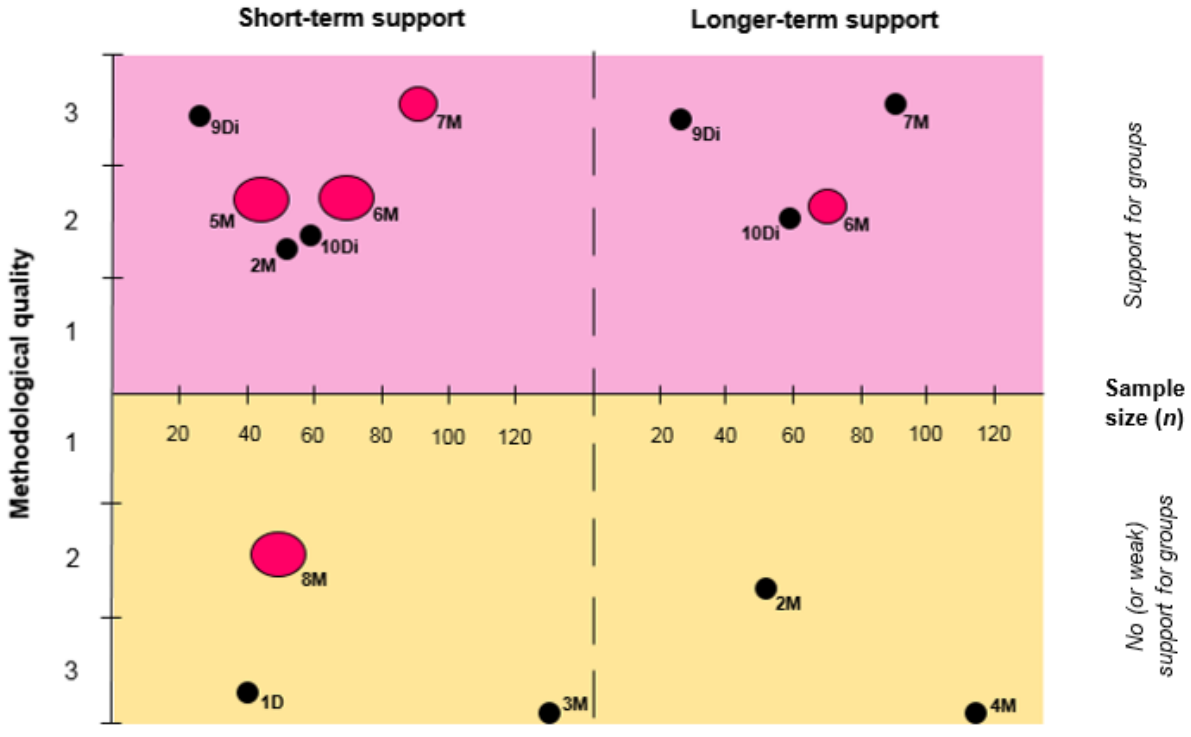
Horn et al. (1987) applied parent and teacher-rated measures of ADHD symptoms, with child-focused measures of locus of control, perceived self-control/concept, academic performance, attention, impulsivity and observed classroom behaviour (e.g. off-task, disruptiveness, etc). The study was designed to compare single (child / parent) versus dual (parent training plus child self-control groups) interventions. Good support was provided for the short and long-term effectiveness of both single interventions. All three groups improved significantly on several child and parent-reported outcomes (hyperactivity, self-control/concept, impulsivity; but not conduct, classroom or academic measures). There was no evidence that combining parent and child groups led to greater treatment effects. Gains were maintained at follow-up on parent measures and two child measures (self-control, attention). Those in the child self-control group demonstrated a significantly greater decrease in hyperactivity compared to the other treatment conditions.

Horn et al. (1990) evaluated similar interventions to Horn et al. (1987), adding school consultation to each condition to create dual (child / parent groups plus school) and multimodal packages (child, parent and school). Parent and teacher-rated outcome measures of ADHD symptoms, behaviour, social-emotional adjustment and self-

control were utilised, as well as child measures of attention, self-control and academic performance. Both dual and multimodal interventions received short and long-term support. Children in all three treatment conditions demonstrated significant improvements at post-test on numerous parent and teacher (not child) measures. No significant treatment group by time interactions were found. At follow-up, gains in all groups were maintained on parent measures only. Children receiving the multimodal treatment improved to the point where, at follow-up, they were no longer significantly different on one self-concept measure to normal controls. One limitation was the possibility of expectancy effects biasing parent-rated outcomes.

Graphical displays of these overall findings are provided, based on the use of ADHD (*see Figure 2*) and non-ADHD (*see Figure 3*) outcome measures.

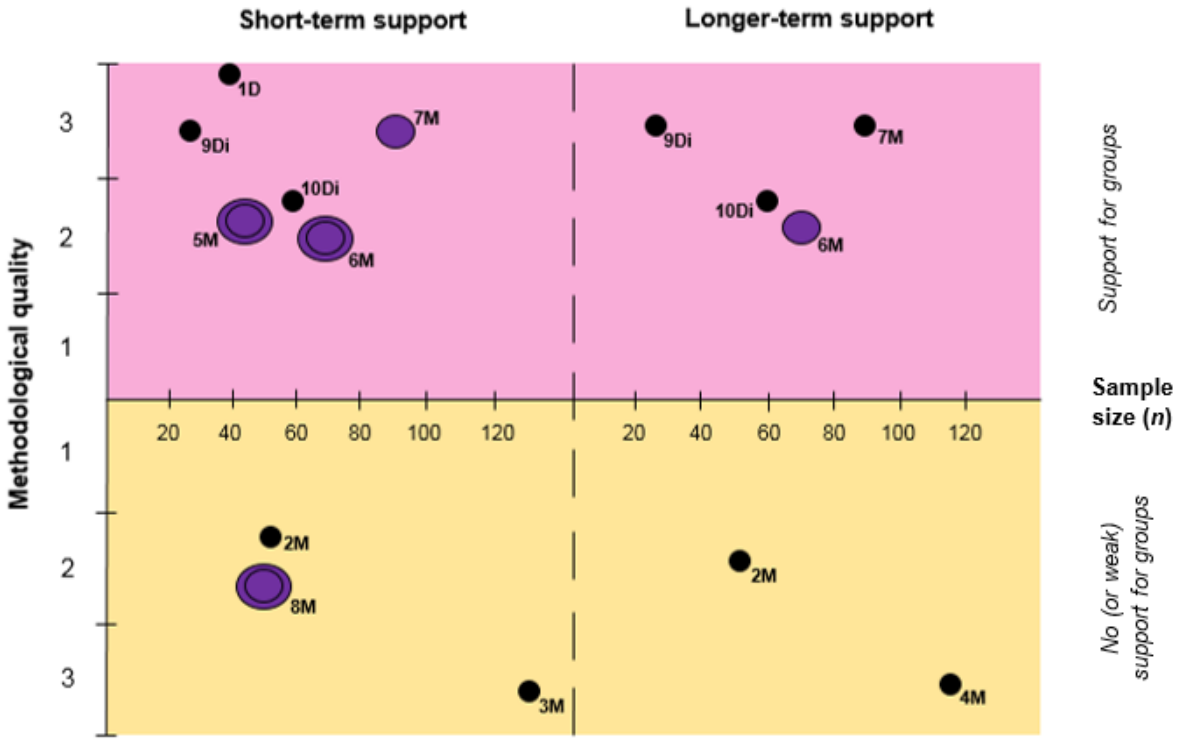
Figure 2: Effectiveness of groups based on ADHD symptom outcomes*



Notes: ● = Effect sizes not reported; ● = Small effect sizes; ● = Medium effect sizes; ● = Large effect sizes. "Short-term" = Pre-post outcomes; "Longer-term" = Follow-up data (any length); Methodological quality as rated by EPHPP; Sample size = Participants originally recruited. Number system: 1 = Fields (2011); 2 = Bloomquist (1991); 3 = Horn (1991); 4 = Ialongo (1993); 5 = Miranda (2011); 6 = Pfiffner (2007); 7 = So (2008); 8 = Van der Oord

(2007) [NB: Effect size is for time, not groups]; 9 = Horn (1987); 10 = Horn (1990). Intervention type: D = Dual, M = Multicomponent, Di = Direct comparison.
** ADHD symptoms were assessed using a variety of measures (neuropsychological tests, rating scales, observations, interviews and questionnaires)*

Figure 3: Effectiveness of groups based on non-ADHD outcome measures*



Notes: ● = Effect sizes not reported; ● = Small effect sizes; ● = Medium effect sizes; ● = Large effect sizes; ● = Medium – Large effect sizes. "Short-term" = Pre-post outcomes; "Longer-term" = Follow-up data (any length); Methodological quality as rated by EPHPP; Sample size = Participants originally recruited. Number system: 1 = Fields (2011); 2 = Bloomquist (1991); 3 = Horn (1991); 4 = Ialongo (1993); 5 = Miranda (2011); 6 = Pfiffner (2007); 7 = So (2008); 8 = Van der Oord (2007) [NB: Effect size is for time, not groups]; 9 = Horn (1987); 10 = Horn (1990). Intervention codes: D = Dual, M = Multicomponent; Di = Direct comparisons.
** Non-ADHD outcome measures included self-concept, executive functioning, externalising/ internalising disorders and so on (see Table 7 for details)*

Discussion

In this review, 22 psychosocial group treatments for children with ADHD, of various study designs, were identified and described. The methodological quality of 19 of these studies was analysed, and subsequently 10 were included in evaluations of treatment effectiveness.

Key findings

Group descriptions

Studies described in this review were nearly all (except one systemic approach) based on behavioural or cognitive-behavioural techniques. Groups focused on a variety of areas, including psycho-education, self-control, problem-solving (usually using self-instructional techniques), mindfulness training, independence and organisational skills, social competence, self-monitoring, anger management, narrative therapy, relaxation and stress management. Groups aimed to improve ADHD symptoms as well as associated difficulties. These approaches seemed feasible and acceptable to most participants. Groups typically facilitated learning through the use of discussions, modelling, guided practice and role plays. Most studies utilised behaviour management techniques and token reward systems, although few implemented response cost procedures. Group size varied but did not exceed ten. Most studies reported group durations of 8 – 12 weeks (range: 3 – 26 weeks) and multimodal approaches were popular. As advised by Kaiser, Hoza and Hurt (2008), the current review paid attention to different levels of approaches (e.g. stand-alone, combined or multimodal treatments). Dual interventions all involved parent training. Multicomponent interventions included parent training, teacher consultation or training (in which daily home-school report card systems were commonly initiated), medication and family sessions.

Sample sizes were generally small. Studies were most often based in the US, although some took place in geographically diverse locations and different cultures. Groups took place in outpatient clinics, schools and (less frequently) laboratory classrooms. Groups were most often delivered by professionals from a psychology background, although some involved paraprofessionals (e.g. nurses, occupational therapists). Participants were mostly aged between 6 to 13 years old. Only 3 of 22 studies included adolescents over the age of 13. Of these studies, two provided

mindfulness training (Van de Weijer-Bergsma et al., 2012; Zylowska et al., 2008) and one had a strong emphasis on academic skills (Sibley et al., 2014). Despite the NICE guidelines (2008) stating that more studies involving adolescent populations were needed, it seems this continues to be significantly lacking in the current evidence base. As is common, samples were predominantly male and White or Caucasian, although some diversity was reported. ODD and CD were the most commonly reported co-morbid disorders. Nearly all studies specified intellectual disorder or IQ < 75 or 80 as an exclusion criterion.

Methodological quality

Child-only groups and dual interventions had the poorest methodological quality, in comparison to multicomponent and direct comparison studies. Poor exploration or control of potential confounders contributed to this in many cases, as did inadequate blinding procedures, data collection methods and poor reporting of drop-out rates. A large number of studies were randomised (16%) or controlled clinical trials (63%), scoring highly for study design. Studies typically did not report information on the rate of acceptance by participants invited to the study, or whether participants were aware of the research question.

Evaluation of effectiveness

Studies were heterogeneous in their aims, use of dependent measures, sample populations (i.e. ADHD subtypes) and interventions. Some studies aimed to reduce ADHD symptoms, while others also targeted treatment at a variety of related difficulties including executive functioning, self-control/concept, social skills and academic performance. No studies with adolescents (13+) were included in the effectiveness analysis due to poor methodological quality or the inclusion of adults in analyses. This was disappointing given the scarcity of outcome data in this area (NICE, 2008).

The effectiveness of child-only groups unfortunately could not be assessed, again due to weak methodology, although one direct comparison study demonstrated some benefits in the short and long-term (Horn et al., 1987). Only one dual intervention (Fields & Hale, 2011; psycho-education) was included in the effectiveness evaluation, of which the results were not strongly supportive of group outcomes in comparison to waitlist controls. Improvement observed in both groups may have been due to expectancy effects or contamination issues, given that all participants continued to receive treatment as usual. Many multicomponent studies found good support for effectiveness in the short to long-term (up to 12 months), observing medium to large effect sizes. Unfortunately many findings were compromised by methodological limitations. Most studies were likely to be underpowered to detect small-medium effect sizes, due to small sample sizes.

Several studies evaluating the benefits of adding multicomponent cognitive-behavioural treatment as an adjunct to medication had mixed outcomes. So, Leung and Hung (2008) found some short-term indications of combined conditions being superior to medication alone. However, at 12 month follow-up combined and methylphenidate-only conditions were equally effective in reducing ADHD symptoms, echoing some findings of the MTA study (MTA Cooperative Group, 1999). Conversely, other studies found little to no support for the superiority of combined behavioural treatments relative to medication alone (Van der Oord et al., 2007; Horn et al., 1991). Although there were indications in Horn et al. (1991) that in some instances, the effects of high dose medication on ADHD symptoms could be achieved by combining low dose with behavioural therapies.

Overall, there were too few direct comparison designs to conclude whether particular levels of approach (single, dual, multicomponent) are more superior or lead to better outcomes. The heterogeneity of studies dictates that in order to dismantle active treatment components it may be best to look at within-study treatment manipulations

(i.e. direct comparison studies) as opposed to between studies, which are difficult to compare. Preliminary evidence from the two studies included in this review had mixed findings. Horn et al. (1987) examined single vs dual approaches and found more support for child-only groups, in the short and long-term. Horn et al. (1990) studied dual vs multicomponent approaches and found equal support in the short-term but slightly more support for multicomponent in the longer-term.

Limitations

Review-level

This review is limited to published, peer-reviewed journal articles in the English language, although positively, many identified studies were from global origins. It was not within the scope of this review to locate unpublished studies or search grey literature, thus study selection may be subject to publication bias. Search strategies were as broad and inclusive as possible, however were subject to the specific terms and restrictions applied. Searches were limited to three databases, which did not include the Education Resources Information Centre (ERIC). This could have enabled further identification of studies within educational settings. Ideally, databases would have been searched from inception as opposed to from 2007, however this was necessary given limited resources.

Identification of studies published before 2007 was dependent upon the search processes of several other systematic review and guideline authors. Reference lists were combined in an attempt to achieve a good coverage of all psychosocial treatment approaches and study designs, however remained limited in the absence of any all-encompassing previous reviews. The present review excluded studies focusing on social skills, ADHD with particular co-morbid conditions and other therapeutic approaches subjectively considered less relevant (e.g. yoga, music/art

therapy). Summer treatment programs were also not included. Lastly, only one author conducted the review process, rendering the study selection process subject to bias.

Study and outcome level

Selected studies involved children aged 5 – 18 years, thus the findings of this review do not apply to children under 5 years or adults therefore limiting generalisability to these age groups. The methodological quality of selected studies has been previously discussed, but one common issue of note in interpreting findings was a reliance on unblinded outcomes, from raters who may have been invested in or had expectations of treatment effects. Sample populations were often unrepresentative and biased towards young white males. ADHD subtypes within sample populations varied but seemed to be biased to some degree towards combined and hyperactive/impulsive types. Many studies had small sample sizes and were most likely underpowered to detect small-medium sized treatment effects.

Conclusions

Group treatments are a feasible and acceptable approach for most children with ADHD and their families. This review aimed to describe and evaluate the effectiveness of psychosocial groups for children with ADHD, which was achieved to some extent but limited by heterogeneous studies with poor methodological quality. It did not succeed in evaluating adolescent interventions, which are particularly sparse in the literature (NICE, 2008). It is clear that further research is needed. Studies provided evidence for various approaches towards the treatment of ADHD, focusing mainly on behavioural or cognitive-behavioural interventions. The broad range of aims, dependent measures and settings of research studies is most likely reflective of the pervasive and multifaceted nature of ADHD.

Implications for practice

Group-delivered interventions are likely to be more cost-effective than individually-administered treatments. They also have additional benefits of peer support for both children and parents. This review provides what are hoped to be helpful descriptions of the variety of group treatment approaches that may be provided, either as stand-alone, dual or multicomponent interventions, to children with ADHD and their families. These findings are relevant for healthcare providers and therapeutic workers based in various paediatric clinical settings or primary and secondary educational contexts. This review promotes the use of group approaches, particularly when embedded within multicomponent treatment packages. These approaches have received positive indications for short and longer-term effectiveness in relation to a diverse range of outcomes, including ADHD symptoms and associated difficulties. Even in cases of limited benefits, no adverse effects of group treatments have been reported. These preliminary findings are likely to also be of interest to policy makers, service-users and their families.

Implications for research and future directions

The evidence base for group-delivered psychosocial approaches for ADHD in children remains limited. More studies explicitly investigating group approaches (preferably comparing these to individually-delivered treatments) are needed, particularly with adolescent populations. Ideally these will involve high-quality RCT designs with blinded raters, which will enable future reviews to better establish the effectiveness of interventions. This particularly applies to child-only and dual interventions, which were frequently limited by weak methodological quality. It would also be helpful to have more direct comparison studies, dismantling treatments to uncover active components. Given that multimodal treatments are more costly it will be important to more clearly establish in what circumstances, treatments and with which populations (i.e. ADHD subtypes, levels of complexity and co-morbidity),

different levels of approaches are most effective. The reporting of effect sizes should be a standard all studies aim to meet, to further allow for comparative analyses.

It is common for studies to exclude children with neurological conditions and learning disorders. Once group approaches receive further support regarding effectiveness, it will be important to consider designing and adapting protocols to meet the needs of these children.

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Part 2: Empirical Paper

Group work for children with Tourette syndrome: A pilot randomised controlled trial evaluating tic severity and neuropsychological outcomes

Abstract

Aims

Tourette Syndrome (TS) is a neurodevelopmental disorder characterised by chronic motor and vocal tics, as well as specific impairments in neuropsychological functioning. Comprehensive Behavioural Intervention for Tics (CBIT: Woods et al., 2008) is a behavioural therapy with strong empirical support for treating tics when individually-delivered. The objective of this exploratory study was to evaluate the effectiveness of newly manualised group-based CBIT, compared with a psycho-educational control group.

Method

This study followed a repeated-measures (pre- to post-intervention), single-blinded randomised and controlled design. Thirty-three children aged 9 to 13 years with TS or a chronic tic disorder were randomised to eight-week CBIT or psycho-educational group treatment. Outcomes of interest were tic severity and neuropsychological functioning (response inhibition, cognitive flexibility and fine motor skill).

Results

An intention-to-treat analysis indicated significant improvements in tic severity for both groups. The medium to large effect sizes observed were comparable to individually-delivered CBIT treatments. Motor tic severity showed greater improvements in the CBIT group ($p = 0.01$, $\eta_p^2 = 0.18$, medium effect), most likely because children primarily chose to treat motor tics (73%) in the CBIT treatment arm. Both groups improved significantly on a test of response inhibition ($p = 0.02$, $\eta_p^2 = 0.20$, medium effect).

Conclusions

There is preliminary evidence to support both CBIT and psycho-education group interventions as feasible and effective, in terms of improving tic severity and inhibitory processes of neuropsychological functioning. CBIT was superior in reducing motor tic severity. Clinical implications and suggestions for future research are discussed.

Introduction

Background

Tourette Syndrome (TS) is a childhood-onset neurodevelopmental disorder characterised by chronic motor and vocal tics. It is thought to affect 0.3-0.8% of school-aged children and adolescents (Scahill et al, 2005; Centers for Disease Control and Prevention: CDC, 2009; Scharf et al., 2012), with an overall international prevalence of 1% (Robertson, Eapen & Cavanna, 2009). Tourette syndrome was first described by Gilles de la Tourette over a century ago. Tics are recurrent, sudden, non-rhythmic motor movements (e.g. eye blinking, jerking of the head, facial grimacing) or vocalizations (e.g. throat clearing, grunting, words) which can vary in complexity. TS is clinically characterised as the occurrence of two or more motor tics and at least one vocal tic, which begin in childhood (<18 years). Tics are likely to fluctuate in frequency ("waxing and waning") but must endure for at least one year following tic onset (Diagnostic and Statistical Manual of Mental Disorders, 5th ed.; DSM-5; American Psychiatric Association, 2013). Symptoms must not be attributable to other medical conditions or the effects of drugs or medication. Persistent (chronic) motor or vocal tic disorders (CTDs) are diagnosed when children experience one or more motor *or* vocal tics, but not both. CTDs may affect up to 2 – 3% of school-aged children (Peterson et al., 2001). This report uses the term "TS" to collectively refer to both TS and CTDs.

Boys are reported to be three to four times more likely to develop TS than girls (McNaught & Mink, 2011). Psychiatric co-morbidity in TS populations are high, with attention-deficit hyperactivity disorder (ADHD) and obsessive-compulsive disorder (OCD) co-occurring most frequently (Zinner & Coffey, 2009).

Tics are often preceded by sensory, or "premonitory" urges (PUs), which have been described as sensations localized to the site of the tic or a general build-up of tension,

which are perceived as aversive and are temporarily relieved on completion of the tic (Steinberg et al, 2010). Unsurprisingly, measures of tic-related PU have demonstrated strong positive correlations with tic severity (Woods et al., 2005).

There is well-established evidence for the neurobiological basis of TS. Anatomical (e.g. Plessen, Bansal & Peterson, 2009) and neuroimaging studies (e.g. Peterson, 2000; Jackson et al., 2011) have indicated dysfunction within the cortico-striato-thalamo-cortical (CSTC) circuits. Wang and colleagues (2011) used functional Magnetic Resonance Imaging (fMRI) to investigate neural activation during spontaneous tics in TS. In brief (see Appendix 5 for further details), it was concluded that tics are generated by a combination of excessive activity in motor pathways (i.e. basal ganglia; motor cortex) and reduced activity in the inhibitory control portions of the CSTC circuits, such as the anterior cingulate and caudate. The level of dysfunction in these circuits was found to be proportionate with tic severity.

Peterson and colleagues (1998) used fMRI to investigate changes in neural activity when adult TS sufferers actively tried to suppress tics. Tic suppression was associated with decreased activity in basal ganglia regions, as well as increased activity in somatosensory, control and attention-related subcortical and cortical brain regions (including the caudate nucleus, anterior cingulate and frontal regions). Neuroplastic changes in control portions of the brain may help to modulate tic severity (Plessen, Bansal & Peterson, 2009). Heightened cognitive control of motor tics is associated with structural and functional changes in the prefrontal cortex (Jackson et al., 2011). Exercising cognitive control mechanisms through the suppression of tics may act to reduce cortical hyper-excitability that gives rise to tics (Jackson et al., 2013).

Researchers have also used behavioural paradigms to demonstrate specific impairments in neuropsychological functioning in TS. In a study of children with TS, Bloch and colleagues (2006) demonstrated that performance on the Purdue

Pegboard test, which assesses fine motor skill, was found to negatively correlate with tic severity at the time of testing as well as predicting tic severity into adulthood. People with TS have also been found to perform poorly on executive functioning measures of response inhibition (e.g. Deckersbach et al., 2006, visuospatial priming [VSP] task; Crawford, Channon & Robertson, 2005, Flanker task) and verbal fluency (Schuerholz et al., 1996). However, researchers are yet to reach a consensus on whether TS is truly associated with executive dysfunction. Conversely other studies have found typical or enhanced levels of cognitive control in children with TS (e.g. Jackson et al., 2007 & 2011). These inconsistent findings have been linked to the influence of co-morbid disorders such as ADHD (Lin, Lai & Gau, 2012), use of a wide range of tests and mixed age samples.

Treatments

There is no known cure for TS. Medication can be effective in reducing tics (Roessner et al., 2013; Weisman et al., 2013), typically by 25 - 50% (Roessner et al., 2011), however side effects often render medication unaccepted or difficult to tolerate (Scahill et al., 2006). The European clinical guidelines for TS (Roessner et al., 2011; Verdellen et al., 2011a) discuss the potential benefits of medication and behavioural treatments, both of which have strong evidence bases. The guidelines do not rate either type of treatment as superior, highlighting the importance of patient choice and symptom severity.

The assumption in behavioural interventions is that tics are involuntary, however can be managed using certain techniques which focus on internal and external factors (*e.g. Habit Reversal Training, see below*). Behavioural approaches aim to interrupt negative reinforcement cycles which may strengthen or maintain the need to tic, which occur when the aversive experience of the PU is temporarily relieved upon completion of a tic (Himle et al., 2007). Behavioural treatments can be used as an adjunct to medication or as an alternative treatment. Habit Reversal Training (HRT)

and Exposure with Response Prevention (ERP) are recommended as first-line treatments, although HRT has the strongest empirical support (Verdellen et al., 2011a; Van de Griendt et al., 2013).

Habit Reversal Training, originally conceptualised by Azrin and Peterson (1988), has demonstrated tic reductions of 30% to 100% (Verdellen et al, 2011a). HRT involves the practice of inhibiting tics through awareness training (recognising the PU and subsequent tic occurrence) and carrying out "competing responses" (CRs) that are physically incompatible with tics, until the PU subsides. Reduction in the aversive PU through *not* completing a tic is thought to reduce the likelihood of a tic re-occurring, through the process of negative reinforcement (Verdellen et al., 2011a). In clinical practice with children HRT also commonly includes concurrently educating parents about TS, in order to reinforce skills learnt and facilitate generalisation to the home environment. HRT has recently been further developed into a Comprehensive Behavioural Intervention for Tics (CBIT: Woods et al., 2008). CBIT is based on HRT techniques and includes additional relaxation training (to reduce anxiety or tension which can worsen tics) and functional-analysis components, the latter of which involves consideration of environmental or external factors in triggering or worsening tics.

There have been two large-scale multisite randomised controlled trials (RCTs) evaluating the efficacy of CBIT delivered on an individual basis. Piacentini and colleagues (2010) found CBIT to be a superior and efficacious intervention for treating tics in children aged 9 to 17 when compared with a psycho-education and supportive therapy control group, with medium effect sizes observed. Benefits were maintained up to six months later in treatment responders, with additional benefits in psychiatric and psychosocial functioning (Woods et al., 2011). Wilhelm and colleagues (2012) similarly found (individually-delivered) CBIT to be a superior intervention for adults with TS compared with a similar control group. Recent meta-analyses have

demonstrated strong support for the effectiveness of HRT and CBIT in reducing tic severity (Wile & Pringsheim, 2013), yielding medium to large effect sizes which are comparable to trials of antipsychotic medication for TS (McGuire et al., 2014). ADHD has been indicated as a moderator for HRT and CBIT treatment outcomes (McGuire et al., 2014), with baseline symptomatology possibly leading to reduced engagement or ability to suppress tics. Higher pre-treatment impairment in response inhibition has also been associated with a reduced positive response to HRT interventions (Deckersbach and colleagues, 2006).

Deckersbach and colleagues (2014) recently investigated neural changes in the basal ganglia and frontal cortex following a 10-week CBIT program for adults (as in Wilhelm et al., 2012). Participants completed a VSP response inhibition task during fMRI scanning pre- and post-intervention. Compared to healthy matched controls, those with TS demonstrated significant pre to post changes in striatal activation. TS participants had higher baseline levels of activation in this region than controls. Following CBIT this reduced to lower levels of activation than in controls. It was concluded that CBIT may facilitate the normalization of dysfunctional neural circuits in adults with TS.

Group-based treatment for children and adolescents with TS remains largely under-researched, with limited empirical testing. All HRT and CBIT interventions to date have been evaluated when delivered on an individual basis only. Psycho-educational treatment groups for children with TS have been described in the literature (Murphy & Heyman, 2007), with the aim of helping young people with TS understand the condition and manage commonly co-occurring difficulties (e.g. anger, OCD, ADHD). These groups are yet to be empirically evaluated and, unlike CBIT, do not focus on tic reduction. Other studies have noted the benefits of psychological interventions focused on improving tic-related impairment and quality of life (McGuire et al., in press), however the treatments were delivered on an individual basis.

Group interventions are promising as they would enable education or interventions to be carried out while simultaneously encouraging peer support and a sharing of experiences (e.g. Tynes et al., 1992). Childrens' learning would most likely be enhanced through social learning processes (Bandura, 1977), by observing modelling by peers and adult facilitators. Groups also have implications for cost-effectiveness and would increase treatment options for patients, both children and parents. Empirical studies of effectiveness would enable families to be fully informed of the expected benefits from different types of groups. Group-delivered treatments for children and adolescents have shown promising indications for effectiveness in other neurodevelopmental disorders (e.g. high-functioning autism spectrum disorders: Reaven et al., 2012; ADHD: Antshel & Remer, 2003). Clinicians and families may have concerns about the possible adverse effects of group treatment, such as exposure to other children with TS resulting in contagion and a worsening of tics ("catching" tics). It would be beneficial to investigate this further and obtain empirical evidence for or against this concern.

Rationale and Aims

There are no known studies investigating tic severity or neuropsychological outcomes for group-based treatments in children and adolescents with TS or CTDs. This exploratory study aimed to examine the effectiveness of CBIT (Piacentini et al, 2010), newly adapted into a group format, in comparison with a psycho-educational group (Murphy & Heyman, 2007).

CBIT aims to help children manage tics by increasing awareness and inhibitory control processes relating to the PU and resulting tics. If children in these groups were to experience improvements in tic severity, it was assumed that this would reflect changes (possibly partial normalization) in underlying neural circuits responsible for tic generation and suppression. A behavioural paradigm, using neuro-behavioural testing, was used as an indirect measure of functioning in these areas.

Specific domains of interest in the present study were response inhibition, cognitive flexibility and fine motor skill.

Whether children with TS would demonstrate change on neuro-behavioural measures after eight weeks of CBIT treatment was unknown, prompting an exploratory approach. The findings by Deckersbach and colleagues (2014) were encouraging. Porto and colleagues (2009) also demonstrated that learning-induced neuroplasticity as a result of cognitive behavioural therapy interventions in adults has been observed in as little as four sessions of treatment.

Hypotheses

Primary (between-groups)

1. Children in the CBIT group will show significant pre- to post-treatment improvements in tic severity and neuropsychological functioning (response inhibition, cognitive flexibility and fine motor dexterity), compared with those in the psycho-educational (PE) group for whom little or no changes are expected.

Secondary (within- and between-groups)

2. Children in the CBIT group will show a significant increase in PU awareness from pre- to post-intervention, compared with those in the PE group for whom little or no changes are expected.

3. a) At baseline, higher levels of neuropsychological functioning (response inhibition, cognitive flexibility, fine motor dexterity, motor-based processing speed) will be correlated with better tic suppression scores and lower tic severity.

   b) Pre- to post-treatment changes on measures of neuropsychological functioning (response inhibition, cognitive flexibility and fine motor dexterity)

will be positively correlated with changes in tic severity and suppression ability *(CBIT group only)*

4. Higher baseline neuropsychological functioning (response inhibition, cognitive flexibility, fine motor dexterity, motor-based processing speed) will correlate with, and predict, greater pre-post improvements in tic severity and suppression ability *(CBIT group only)*

5. Lower baseline ADHD symptomatology will correlate with, and predict, greater pre-post improvements in tic severity and suppression ability *(CBIT group only)*

Method

Design

This study followed a repeated-measures (pre- to post-intervention), single-blinded randomised and controlled design. Children were randomised to either the experimental group (CBIT) or the control group (PE). A parallel design was employed which included four group-based parent workshops. The parent groups were run concurrently whilst children were attending the groups.

The wider project

This study was part of a wider joint project with Rachel Yates, a Trainee Clinical Psychologist from Royal Holloway, University of London (RHUL). The author and this trainee are referred to in this report as “the researchers”. These projects both investigated the effectiveness of the two group treatments in terms of tic severity outcomes, however they each had different research questions. The present study focused on neuropsychological functioning, whereas the other project explored quality of life outcomes (Yates, 2014). A complete outline of both trainees’ contributions to the joint study is provided in Appendix 6.

Child and parent satisfaction questionnaires *(*see Appendices 7 and 8*)* from the groups were analysed by a placement student to fulfil the academic requirements of a Masters qualification (İnce, 2014). All measures that were administered as part of the wider study are outlined in Appendix 9.

Participants

Eligibility criteria

The eligibility criteria for this study are outlined in Table 1. The age range of participants was carefully considered. TS symptoms are well-documented as reaching their peak of severity at 10 - 12 years old (McNaught & Mink, 2011; Cutler et al, 2009). To increase recruitment opportunities this age range was expanded to include 9 - 13 year olds. This approach fitted well with new referrals to the TS clinic, in which children were on average aged 11 years old.

Table 1: Inclusion and exclusion criteria

Inclusion Criteria	Exclusion Criteria
Children with diagnoses of TS, Chronic Multiple Tic Disorder (CMTD) or Chronic Vocal Tic Disorder (CVTD)	Full-scale IQ <80 (as in Piacentini et al., 2010)
Males and females	Previous or current psychosis, substance abuse or dependence
Aged 9 - 13 years old	Tics not considered the primary disorder
Co-morbid disorders (e.g. OCD, ADHD) were accepted as long as tics were considered the primary disorder.	Children who had previously received more than four sessions of HRT or CBIT
Families were required to have a good enough use of English to be able to follow sessions.	Children who had attended the psycho-educational group within the last two years
[To reduce travel commitments for families and researchers, catchment areas within an hour of London were prioritised, but this was not essential]	Baseline score of <13 on the Yale Global Tic Severity Scale (YGTSS; Leckman et al., 1989)

Setting

Participant recruitment and the running of the groups took place in a national TS outpatient clinic based within a children's hospital. Families were recruited through new referrals and retrospectively, inviting children who had been assessed or treated at the clinic within the past five years.

Ethical approval and funding

The ethics and practicalities of this study were reviewed and approved by University College London (UCL), RHUL and the hospital's Clinical Research Adoption Committee (see Appendices 10 to 12). It was then reviewed and approved by the London Queen Square Research Ethics Committee (see Appendices 13 to 14). The study was registered with an International Standard Randomised Controlled Trial Number (ISRCTN; 50798741) via the National Institute for Health Research (NIHR) Portfolio Database (http://www.controlled-trials.com). Funding was provided by UCL, RHUL and Tourettes Action (registered charity 1003317; see Appendix 15). Unfortunately there was no funding to contribute towards families' travel costs, although funding was used to award four £50 Amazon vouchers, one at the end of each group (2 x CBIT; 2 x PE). Children were eligible to receive this if they had attended six sessions or more. Vouchers were awarded to eligible children who had shown the best behaviour in each of the groups, as demonstrated on a behaviour monitoring chart.

Interventions

Both group treatments were highly structured and manualised (full manuals available upon request; examples of session two of CBIT and PE child groups are provided in Appendices 16 to 17). The treatments were made as comparable as possible in terms of structure, therapist exposure, peer support, use of reward systems and the amount of assigned homework between sessions. Both consisted of eight group sessions on a weekly basis, with the CBIT and PE groups being run in parallel on different days of

the week. The first two sessions were 90 minutes long and the remaining sessions were 70 minutes long. All groups started at 4.30pm and were not run during school holidays. Sessions contained a mixture of didactic teaching, large and small-group discussions and activities. Both types of groups were made as interactive and interesting as possible, to maximise participation.

Both treatment arms received brief psycho-education about tics and were taught relaxation techniques. Reward systems in both groups were the same, to maximise motivation during the sessions and when completing tasks at home. Children were able to gain stars during the groups for desired behaviours such as listening well, sharing their ideas and completing homework. Each child also developed a personalised reward chart with their parents.

Staff for both groups were kept as consistent as possible. The lead facilitator, TM, is a Consultant Clinical Psychologist and Paediatric Neuropsychologist with over ten years of specialist experience in the national TS clinic. TM led all child groups (CBIT and PE), supported by two to three facilitators of various professions (e.g. specialist nurses, clinical / trainee / assistant psychologists). Treatment fidelity was measured every session for both groups by an assistant psychologist, using the manual as a checklist. The assistant provided direct prompts to therapists during sessions if any content had been missed.

CBIT groups

The manual for group-based CBIT was primarily developed by TM, with some input from other colleagues. The CBIT groups combined and adapted protocols outlined in an HRT treatment workbook (Verdellen et al., 2011b) and manual (Woods et al., 2008) for children and adolescents with TS, which were both originally devised for one-to-one treatment.

The CBIT group sessions followed a similar structure each week: Welcome, review homework, rate tic severity for the past week, one long activity, break, one short activity, homework setting and reuniting with parents. CBIT session content included: Brief psycho-education on TS and CBIT / HRT, tic awareness training, functional analysis of tics, weekly self-rating of tic severity, developing and practicing competing responses for the three most severe or impairing tics (see Appendix 18 for examples; 2 - 3 children only worked on one or two tics if they were still struggling to consolidate the effectiveness of the technique), progressive muscle relaxation and relapse prevention. It is common within individual and group CBIT sessions to only focus on the most severe tics, in order to teach children skills which they can spend time mastering with support, to later apply to any other tics should they wish.

Psycho-educational groups

The PE group manual followed those that had been run at GOSH (Murphy & Heyman, 2007), but was extended by adding two sessions to the protocol to make it equivalent in length to the CBIT group.

The PE group sessions followed a similar structure to CBIT each week, with the omission of rating tic severity each week. PE sessions primarily followed a cognitive-behavioural model. The sessions covered topics on TS and common co-morbid difficulties including: Psycho-education on TS and treatments (e.g. HRT; medication), self-esteem, school and bullying, anxiety and OCD, planning and organising, anger (including progressive muscle relaxation) and attention. The final session included a quiz and a consolidation of topics.

Parent groups

The aim of parent groups was to provide psycho-education about tic disorders, followed by specific information on topics that were covered in the allocated child groups. This parallel approach was to facilitate generalisation of skills learnt. Involving

parents in their child's treatment is commonly reported in studies of group interventions for children with neurodevelopmental disorders (e.g. Fields & Hale, 2011; Van de Weijer-Bergsma et al., 2012). There have been indications that active parental involvement enhances the outcomes of group therapy for children with neurodevelopmental conditions (e.g. Sofronoff, Attwood & Hinton, 2005).

Parent groups were fully manualised (developed by TM and the clinical team) and were facilitated by two Clinical Psychologists. Parent group sessions only ran during sessions 1 - 4. For the remaining sessions 4 – 8, parents were given a room to wait in together and socialise, with tea and coffee being provided. A Clinical Psychologist was available at the start of these later sessions to answer any questions that had arisen that week and collect the homework.

Each week parents in both groups were asked to subtly observe and count their child's tics for 15 minutes every day at home ("tic tracking"), using a silent counter and recording the data on a sheet provided. This was as described in the HRT clinician guide for children (Verdellen et al., 2011b), providing an estimate of tic severity and weekly progress in-between sessions.

Additional aspects of treatment packages

All children received medication management[2] and school liaison work by the clinical team irrespective of group assignment, as part of routine care. While taking part in the study children did not receive any other forms of psychological treatment within the team at the clinic, although a small number of children received input from other professionals externally (as reported in *Results*).

Outcome measures

[2] Children were allowed to be on medication but families were requested not to change medication dose throughout the study.

Measures for the current study only are reported. For a full list of all measures administered at pre- and post-assessment, including those for the wider study, see Appendix 19. These are also detailed in the pre- and post-assessment protocols later described (see Appendices 20 and 21).

Pre-assessment only

- **Demographics questionnaire**

This was a structured form (see Appendix 22) followed by researchers when initially contacting families over the telephone. It gathered information relating to exclusion criteria, co-morbid disorders, ethnic background[3], previous and current treatment and socioeconomic status (SES) of the family. SES was calculated using information relating to parental education and occupation (Hollingshead, 1975).

- **OCD**: **Children's Obsessional Compulsive Inventory - Revised** (**CHOCI-R:** Uher et al., 2008)

This is a 32-item self-report questionnaire, with both child-rated and parent-rated scales, to assess symptoms of OCD-related symptoms and impairment.

- **ADHD: The Swanson, Nolan and Pelham Questionnaire** (**SNAP-IV, MTA version:** Swanson et al., 2001)

This is a 26-item self-report measure designed for parents and other caregivers, which assesses for symptoms of ADHD. Separate subscale scores are provided for ADHD-Inattentive, ADHD-Hyperactive/Impulsive and combined subtypes.

- **IQ: Wechsler Intelligence Scales for Children, 4th edition, short form** (**WISC-IV-SF**: Crawford et al., 2010)

[3] Data regarding ethnicity was later entered according to particular categories, as used in the 2011 Census codes and advised by the Office of National Statistics (2011).

This measure was primarily used to ensure children met eligibility criteria for the study. The WISC coding subscale was also used as a measure of motor-based processing speed for hypotheses involving baseline neuropsychological functioning.

Pre- and post-assessment

- **Yale Global Tic Severity Scale (YGTSS; Leckman et al., 1989)**

The YGTSS was the primary outcome measure for this study and is widely considered the gold-standard measure of tic severity. This is a well-validated clinician-rated instrument which is based on semi-structured clinical interviews with children and parents.

Clinicians review a list of all possible motor and phonic tics that the child may have experienced over the past week. The clinician then evaluates (each on a 5-point Likert scale) the number, frequency, intensity and complexity of tics that week, as well as how much they interfere in daily life. This is evaluated separately for motor and phonic tics (each subdomain is scored from 0 - 25). Children and parents are then also asked about the overall impact of tics on their life ("impairment", 0 to 50), which is combined with the motor and phonic subscales to form a total score, ranging from 0 to 100.

The YGTSS has demonstrated good construct, convergent and discriminant validity, as well as good internal consistency and test-retest reliability, in child and adult populations (Leckman et al., 1989; Storch et al., 2005).

YGTSS interviews were filmed (with permission) and 20% of videos were re-scored for inter-rater reliability.

- **Premonitory Urge for Tics Scale** (PUTS; Woods et al., 2005)

This brief, 9-item self-report scale is designed to provide a measure of tic-related premonitory urge awareness in children and adolescents (e.g. "Right before I do a tic,

I feel pressure inside my brain or body"). It uses a 4-point Likert scale, giving total scores ranging from 0 to 36. The PUTS has demonstrated good criterion validity (significant correlations with the YGTSS and other related measures), internal consistency (α = 0.81) and test-retest reliability (Woods et al., 2005). In this study, the PUTS demonstrated good internal consistency (α = 0.874 – 0.889, pre- and post-assessment respectively).

- **Direct observation of tic expression and tic suppression** (Based on the protocol reported by Himle et al., 2006)

Direct measures of tic behaviours are recommended to supplement indirect measures such as the YGTSS (Himle et al., 2006), which rely on retrospective reporting. Some studies have found no significant correlations between these types of measures (Himle et al., 2006), indicating they are likely to measure unique aspects of tic severity. Direct measures focus on the frequency of tic expression, whereas the YGTSS also takes into account tic strength, complexity, interference and impairment.

Children were filmed for 15 minutes (tics observed) and then 5 minutes (suppression task), whilst sitting at a table and watching a pre-defined DVD (The Simpsons). Careful consideration was given regarding which Simpsons episodes to show at pre- and post-assessment. Details of the DVD selection process are described in Appendix 23. In the suppression task, children were asked to continue watching the DVD but to suppress ("hold in") any tics. They were told they would receive a small reward for this at the end (a stretchy man at pre-assessment or slinky toy at post-assessment, to ensure the same novelty effect).

A clear protocol was followed (see Appendices 20 and 21 for pre- and post-assessment protocols). This protocol was based strongly on that of Himle and colleagues (2006), which has demonstrated good test-retest reliability (Piacentini et al., 2006). An exception was that filming was conducted using in-built webcam

software on a laptop, instead of a video camera. Unfortunately this meant that only the upper torso and head were visible for filming. The filming periods of 15 minutes and 5 minutes were based on practicalities and research by Himle and colleagues (2006), who investigated and advised optimal observational lengths.

Scoring

A Clinical Psychologist and a Masters-level placement student ("raters") scored all videos. The raters were instructed to follow a detailed scoring protocol (see Appendix 24), with the use of a scoring sheet (see Appendix 25). For full details of the scoring process, see Appendix 26. In brief, raters counted the number of tics seen to generate a total tics per minute index score, for observation and suppression conditions. Inter-rater reliability with "expert" raters (the researchers) was calculated for 20% of the videos.

- **Computerised measures of neuropsychological functioning (NIH Toolbox)**

The National Institutes of Health (NIH) provide free access to a variety of brief, online computerised tests of neuropsychological functioning (www.nihtoolbox.org). The tests are standardized, nationally normed and validated across the lifespan, for ages 3 to 85 years. For all selected measures, children were seated at a table and standardized instructions were provided on the screen for researchers to read aloud. Age-adjusted standardised scores were used in the present study for all three tests, in which higher scores indicated better functioning. All timed tasks were measured to the accuracy of 100th of a second. Scores were automatically calculated by the online system. The following measures were administered:

- **Dimensional Card Sort Test (DCST) – Attention, cognitive flexibility and response inhibition (NIH Toolbox)**

This measure took four minutes to administer. Participants were presented with a target picture in the middle of the screen. They were then shown two pictures below, one that matched the colour of the target picture and one that matched the shape. In the first and second rounds, participants were asked to match based on shape and then colour respectively. In the third round, children were required to switch to shape or colour selection rules at random according to the computer's instructions. Each round consisted of practice and test sections. Scores were calculated by the computer based on accuracy and reaction time.

- **Flanker Inhibitory Control and Attention Test – Attention and response inhibition (NIH Toolbox)**

This measure took three minutes to administer. Participants were required to focus on a row of five horizontal arrows, selecting the direction of the middle arrow only using the cursor keys. The surrounding "flanker" arrows were either congruent or incongruent with the direction of the middle arrow. Scores were calculated based on accuracy and reaction time.

- **Motor Dexterity Pegtest (NIH Toolbox)**

This measure of fine motor dexterity took approximately four minutes to administer. A plastic pegboard with nine holes (3 x 3 rows) was secured in front of the child. It was explained that the aim was to accurately place all nine pegs into the holes and then remove them one at a time as quickly as possible. Participants were allowed one practice trial before they were timed using an online stopwatch. Both hands were tested separately, although normative data was available for the dominant hand only.

Procedure

Children were invited to participate in the study via an invitation letter (see Appendix 27), which was posted to retrospective patients or given in person by the researchers or clinical team to new referrals. Along with the invitation letter, each family invited to

participate were provided with information sheets. One was designed for parents or carers (see Appendix 28) and one for children (see Appendix 29). CBIT was not framed as a superior treatment. Both groups were described as active interventions with potential benefits, which group facilitators and researchers endorsed.

Once parents had provided verbal consent for their child to participate in the study, over the telephone or in person, the demographic questionnaire was completed. This was administered over the telephone (*see Outcome Measures*) and arrangements were made for the pre-assessment. The two researchers divided all assessments between themselves. Nearly all pre- and post-assessments were conducted as home visits for the families' convenience[4].

Pre-assessment

Researchers followed a detailed and highly structured pre-assessment protocol (see Appendix 20), administering a battery of tests (also listed in Outcome Measures and summarised in Appendix 19) taking approximately three hours. Parent consent and child assent forms were completed (see Appendices 30 and 31), first ensuring that all information had been understood.

The protocol was carefully designed to be maximally engaging to children by providing a mixture of tasks, whilst also being practical for researchers and families. Visual timetables were provided (see Appendix 32) to give children a clear structure for the day. Regular breaks were provided.

Once children had completed their first assessment visit, they were randomised to a treatment group (see *Randomisation* below). Parents were asked not to change their child's medication status or dosage during the study. Letters were sent to each child's GP to inform them of their participation in the group program (see Appendix 33).

[4] Four pre-assessments were carried out at the clinic, however this was discontinued due to issues with accessing the internet. Direct observations of tics in clinical contexts have been found to correspond well to home-based observations (Himle et al., 2006) and thus were not analysed separately in this study.

Post-assessment

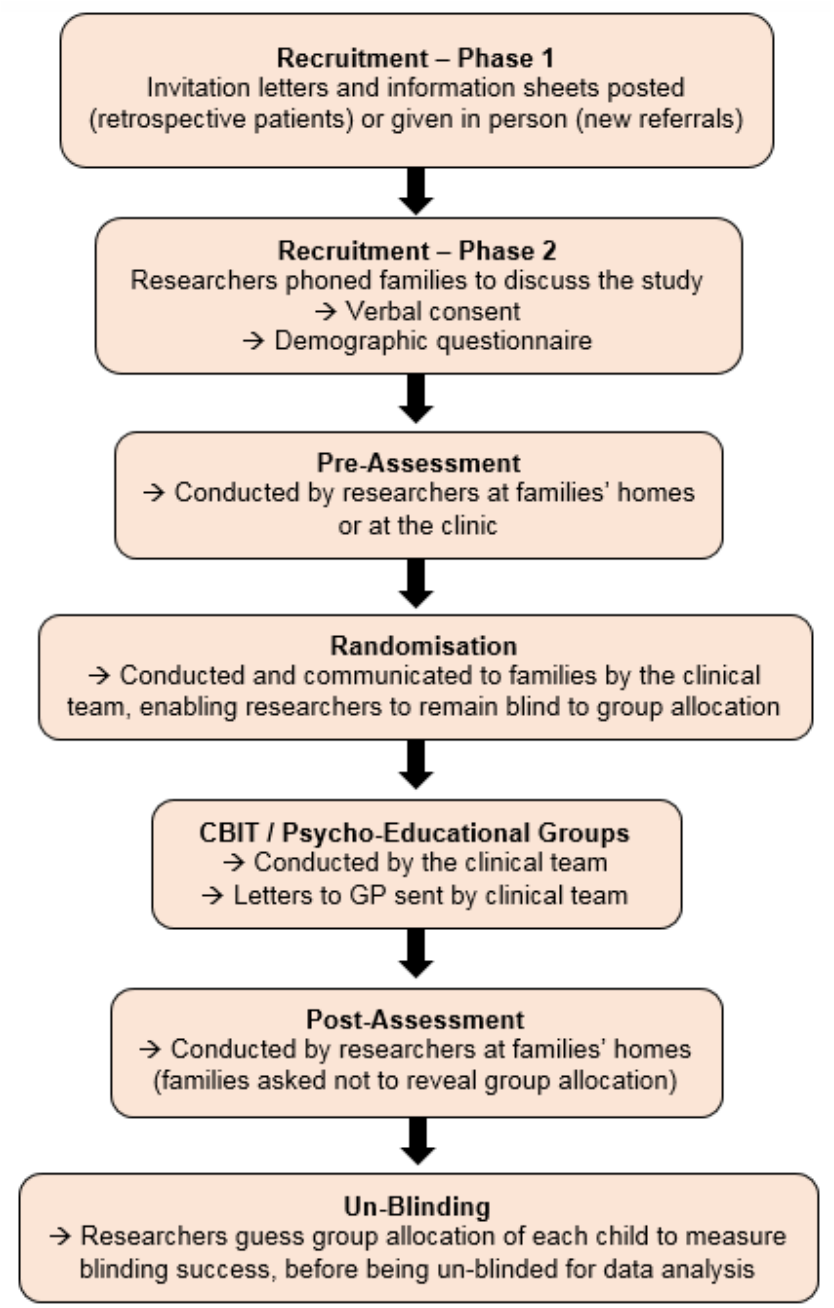
Following the groups, families were again visited at home for a post-assessment lasting roughly one and a half hours. Researchers were kept consistent for pre- and post-assessments for each family to maintain rapport and to facilitate reliability of measurement. This again followed a highly structured protocol, administering measures in the same order as in the pre-assessment. The post-assessment was shorter than the pre-assessment, as several measures were given at pre-assessment only (see *Outcome Measures* and Appendix 19). At the end of the post-assessment parents were asked the questions at the end of the demographic questionnaire, in order to gather information regarding changes in medication, significant life events and to arrange practicalities (see Appendix 22).

The procedures followed in this study, focusing particularly on the process of achieving a single-blinded design, are depicted in Figure 1.

Randomisation

Children were randomised to either CBIT or PE groups using QMinim online minimisation software (http://qminim.saghaei.net/index.php). The minimisation method of randomisation was used to balance the two groups not only by treatment type, but also by age and gender. Children were randomised sequentially as they were recruited, to ensure groups could start as soon as possible.

Figure 1: Flow chart of study procedures and the single-blinding process



Blinding

Researchers were not involved in the randomisation process or running of the groups, in an attempt to remain blind to treatment allocation and reduce the likelihood of bias. A Clinical Psychologist on the team, who was involved in the running of the groups but not in recruitment or assessment, was responsible for randomising each child to a treatment group. This was done using instructions provided by the researchers (see

Appendix 34). The results of randomisation (study ID number and group allocation) were stored on a password-protected file, which the researchers were unable to access. Families were contacted by an assistant psychologist on the team. It was not possible to conceal time point from the researchers, given they were solely responsible for carrying out all assessments. It was also not possible to blind families to treatment allocation due to the overt nature of the interventions. Researchers remained blind to treatment allocation until data was due to be analysed, at which point the success of blinding was assessed (*see Results*).

Direct tic observation video-recorded measures were scored by two raters (*see Measures*) who were blind to treatment allocation and time point.

Practical and ethical considerations

All electronic and paper-based confidential data was stored securely according to NHS and local Trust guidelines. All participants were anonymised with a study ID number. During assessment visits, researchers followed NHS and local Trust Lone Working Policies. See Appendix 35 for further details of practical and ethical considerations.

Service-user involvement

One mother of a child with TS, who was not taking part in the study, was consulted over the telephone (having given permission), with regards to the structure and content of invitation letters, information sheets and consent forms given to parents and children. Overall, feedback was very positive and minor changes were suggested and made. The mother and her son also suggested that a video of The Simpsons would be age-appropriate for the tic observation task, which was subsequently implemented. For full details, see Appendix 36.

At the end of the study, all participating families received a written feedback report which thanked them for their involvement and gave a summary of the main findings of both doctoral projects.

Power calculations

The "G*Power3" computer program (Faul et al., 2007) was used to compute calculations of required sample size. No previous studies were identified which could provide accurate estimates of expected effect sizes for TS group-based treatments for children, in terms of tic severity and neuropsychological outcomes. Calculations were based on a medium effect size (for Repeated-Measures ANOVA, pre-test versus post-test contrast) of f=0.25, which was of most clinical interest. Alpha was set at 0.05, correlation among repeated-measures at 0.5 and nonsphericity correction at 1. Power was set at 0.8. Assuming equal group sizes, with two groups and two primary outcome measures (pre and post YGTSS tic severity), the analysis recommended a minimal total sample size of n = 34. This was acknowledged to be a very tentative power analysis, which is often inevitable when attempting to explore the effects of novel treatments. Presuming drop-out rates of 5% to 27%, as indicated in previous studies of group-based treatments (Himle et al., 2003; Silverman et al., 1999), a conservative aim of recruiting 48 participants (24 per condition; 12 per group) was agreed.

Statistical Analysis

Data was analysed with SPSS 22.0, using three different analytic approaches:

1. An intention-to-treat analysis (ITT), using last observation carried forward for participants lost to follow-up. This was the primary analytic approach.

2. Attenders-only analysis, in which participants who attended five sessions or less were excluded in order to examine outcomes when the protocol was adhered to.

3. Removal of potentially confounding cases: Those who had experienced significant life events or deterioration in mental health during the study (*n* = 3)[5], changes in medication (*n* = 5), receipt of additional current psychological input (*n* = 4); had a significant delay in post-assessment (*n* = 2), or were prescribed stimulant medication at any point during the study (*n* = 1)[6].

Prior to analysing the data, the overall success of blinding was assessed using the chi-square test (2 x 2 contingency table). Inter-rater reliability on the YGTSS and tic observation measures was evaluated using Intraclass Correlation Coefficients.

Descriptive data was then explored and tests of normality conducted. Statistical outliers were removed and transformations were performed when necessary, in an attempt to normalise the data. Parametric tests were used throughout the analyses. Baseline demographic and clinical characteristics between both groups were tested using independent-samples *t*-tests for continuous data and Fisher's Exact tests for categorical data.

Primary comparisons were investigated using 2 x 2 mixed model Repeated Measures Analysis of Variance (RM-ANOVA)[7] tests. Main effects of time (within subjects: pre-post treatment) and group type (between-subjects: CBIT versus PE) were examined, along with group-by-time interactions. Bonferroni-adjusted significance levels were reported to control for inflated Type I error rates from multiple comparisons. Partial

[5] One child was being bullied and had a grandparent who was very ill. One had experienced the birth of a baby brother. One displayed unexplained medical symptoms following the group but before the post-assessment.

[6] Stimulant-related improvements have been documented for a range of cognitive functions such as attention (e.g. Swanson, Baler & Volkow, 2011). Medication status in itself was not analysed as a potential covariate or moderator of treatment effects, as previous studies of behavioural interventions for TS have found no evidence for this (Deckersbach et al., 2006; Piacentini et al., 2010; McGuire et al., 2014).

[7] While it is acknowledged that Multivariate ANOVA (MANOVA) can protect against inflated Type I error rates, it can also provide ambiguous results with no clear indication of which variables are influencing significant findings. Thus, the decision was made to use multiple ANOVA analyses.

eta-squared (η_p^2) effect sizes[8], as calculated through SPSS when conducting an RM-ANOVA, were reported. Age has been found to be a moderator of behavioural treatment effects (McGuire et al., 2014). An Analysis of Covariance (ANCOVA) model was considered, to control for age as a covariate should significant correlations be found with any dependent variables; however no such correlations were found.

Secondary hypotheses were investigated using RM-ANOVA and Pearson's correlations. Pearson's correlation coefficient (*r*) was used as a measure of effect size[9]. In cases where significant correlations were found and it was appropriate given the hypothesis, simple linear regressions were conducted to assess predictive relationships.

Results

The flow of all participants through the study is outlined in Figure 2. Recruitment took place from June to November 2013, with a total of 153 children being assessed for eligibility. Thirty-three children were subsequently recruited and assessed prior to being randomised to either the CBIT (*n* = 17) or PE (*n* = 16) groups.

The first groups (1 x CBIT; 1 x PE) were run from September to November 2013, with a one-week mid-treatment break for half-term. The second round of groups (1 x CBIT; 1 x PE) were run from November 2013 to January 2014, with a 3-week break over the Christmas holidays. All pre- and post-assessments were carried out within one month of a child starting or finishing the group[10]. Each child remained in their assigned group. Data collection was completed by March 2014.

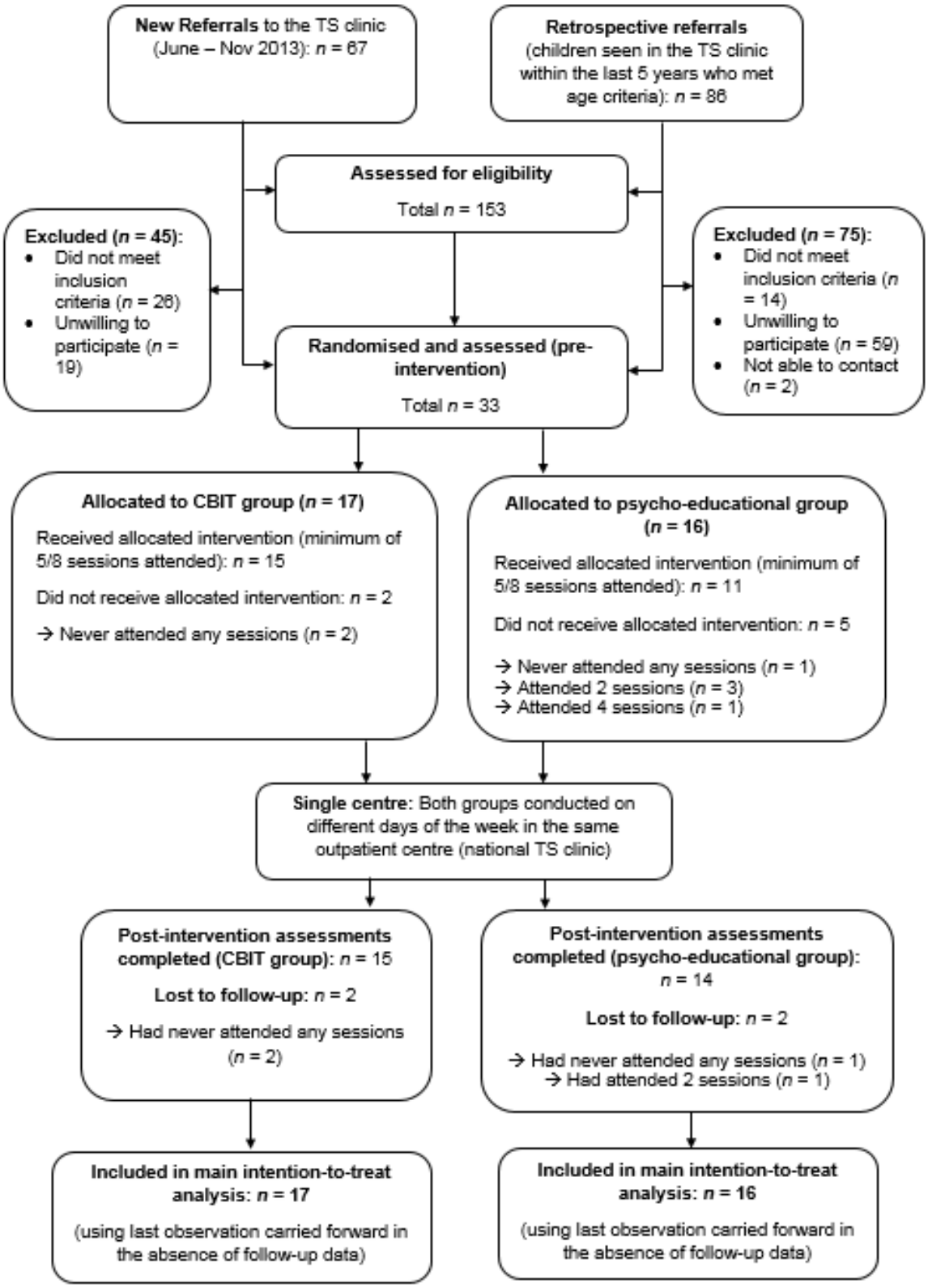
[8] According to rules of thumb interpretations provided by Cohen (1988) and Miles and Shevlin (2001), in which 0.02 – 0.12 represents a small effect size, 0.13 – 0.25 a medium effect size and ≥0.26 a large effect size.

[9] Cohen's (1988) rules of thumb allowed interpretations of magnitude (0.1 to 0.3 = small effect size, 0.3 to 0.5 = medium effect size, ≥0.5 = large effect size).

[10] With the exception of two children, for whom post-assessment was delayed by up to 16 weeks.

Regular attendance to the groups was maintained but better within the CBIT group. 88% of children from the CBIT group attended at least five sessions or more, compared with 69% in the PE group. Four children were lost to follow-up, with three (9%) not attending any sessions at all.

Figure 2: Flow chart of participants' progression through the study



Notes. Diagram presented in line with CONSORT guidelines for non-pharmacological randomised controlled trials (Boutron et al., 2008). See Appendix 37 for a full list of reasons for exclusion and Appendix 38 for reasons families declined to participate.

Generalizability of the sample

In total, 30% of families who were considered eligible and invited to take part in the study accepted. Only 16% of families invited retrospectively accepted invitations to participate, compared with 54% of new referrals. The final recruited sample (*n* = 33) consisted of 76% males and 24% females. The mean age was 10.96 years old. Ethical approval was granted to allow researchers to record reasons that families declined participation in the study, along with the child's age and gender. This was to facilitate an estimation of whether the final recruited sample was generalizable, in relation to all participants who were invited to the study. See Appendix 38 for reasons families declined to participate. Distance to travel was the most frequent reason given for not wishing to participate. Of those invited retrospectively (*n* = 86), 81% were male, with a mean age of 11.03 years old. Of the new referrals (*n* = 67), 73% were male, with a mean age of 11.18 years old. Thus it was concluded that the final recruited sample was approximately representative according to these variables.

Success of blinding

When conducting RCTs it is vital to assess and report the success of blinding (Hróbjartsson et al., 2007). The researchers who had conducted all pre- and post-assessments were required to guess the group allocation of children they had tested. The outcome of these guesses in comparison to actual treatment groups is displayed in Table 2. Overall, blinding was successful within this study ($X^2 = 0.04$, $p = 0.85$). The success of blinding for researchers rating the tic counting measures was not tested.

Table 2: Assessment of the success of researcher blinding to treatment group

		Researcher's guess		
		CBIT	Psycho-educational	Total *n*
Actual treatment group	CBIT	9 (53%)	8 (47%)	17
	Psycho-educational	9 (56%)	7 (44%)	16

Inter-rater reliability

YGTSS

The YGTSS interview videos of 12 participants[11] (20%) were selected for re-coding by a Clinical Psychologist colleague, who was trained in the YGTSS to the same level as the researchers. An online random numbers generator (http://www.random.org/) was used to ensure unbiased random selection. Inter-rater reliability between the original researcher ratings and the re-coded scores was then calculated through Intraclass Correlation Coefficient (ICC). A two-way random effects model with an absolute agreement definition was used.

For the motor tics subscale, a good degree of inter-rater reliability was found. The average measure ICC (2,*k*) was 0.811, with a 95% confidence interval from 0.149 to 0.950 ($F(11,11) = 8.245$, $p = 0.001$). However, the wide and low ranging confidence interval observed indicates poorer reliability than estimated in the average measure value. This implies that inter-rater reliability may not be at an acceptable level.

For the phonic tics subscale, an excellent degree of inter-rater reliability was achieved. The average measure ICC (2,*k*) was 0.939, with a 95% confidence interval from 0.774 to 0.983 ($F(11,11) = 19.635$, $p = 0.000$).

For the Total score (with impairment), an excellent degree of inter-rater reliability was achieved. The average measure ICC (2,*k*) was 0.928, with a 95% confidence interval from 0.582 to 0.982 ($F(11,11) = 21.924$, $p = 0.000$). However, the low range confidence interval observed indicates poorer reliability than estimated in the average measure value. This implies that inter-rater reliability may not have been at an acceptable level.

Direct tic observations

[11] Half of the participants were selected randomly from those originally scored by the first coder, and half from the second coder. Equal numbers of videos were drawn according to time point (pre or post) and date of group commencement (September or November).

24 videos[12] (20%) were selected, with the researchers ("expert" raters) re-scoring half of the videos each. An online random numbers generator (http://www.random.org/) was used to ensure unbiased random selection. Inter-rater reliability between the original raters and the expert raters was then calculated through Intraclass Correlation Coefficient (ICC), using a two-way random effects model with an absolute agreement definition. An acceptable degree of inter-rater reliability was found. The average measure ICC (2, *k*) was 0.771 with a 95% confidence interval from 0.465 to 0.901 ($F(23,23) = 4.240$, $p = 0.000$). However, the wide and low ranging confidence interval observed indicates poorer reliability than estimated in the average measure value.

Treatment fidelity

100% fidelity to the manuals was achieved for both the CBIT and PE groups. The exception to this was that for both groups several sessions ran out of time in the last few minutes.

Normality, descriptive data and baseline group differences

Upon inspection of the data, all variables were found to be normally distributed and the assumptions for parametric testing met. One exception to this was post-intervention data for the tic observation and suppression measure, which was not normally distributed as indicated by Kolmogorov-Smirnov tests ($D(14) = 0.314$, $p = 0.001$ and $D(14) = 0.273$, $p = 0.006$, respectively) and had evidence of positive skew and kurtosis. This was normalised following the removal of a statistical outlier (participant #2, PE group).

[12] 6 participants, 4 videos each: Tic observation and suppression conditions at pre- and post-assessment. Half of the participants were selected randomly from the pool of participants originally scored by the first rater, and half from the second rater.

Continuous and categorical descriptive data from the groups at baseline are presented in Tables 3 and 4 respectively, along with tests of significance. No significant differences were found between groups on any variable.

Table 3: Continuous descriptive data at baseline with tests for group differences

		CBIT group (n = 17)	Psych-Ed group (n = 16)	Total sample (n = 33)	Independent-samples t-test[a]
		M (SD)	M (SD)	M (SD)	p-value*
Age (years)		10.87 (1.31)	11.05 (1.62)	10.96 (1.45)	0.73
Full-scale IQ		100.65 (11.55)	103.13 (13.75)	101.81 (12.48)	0.58
SES total score		38.19 (13.82)	45.91 (14.76)	41.93 (14.60)	0.13
YGTSS	Motor	17.65 (4.74)	16.31 (3.03)	17.00 (4.00)	0.34
	Phonic	12.71 (6.99)	12.63 (5.93)	12.67 (6.40)	0.97
	Total	54.47 (16.43)	50.19 (14.48)	52.39 (15.43)	0.43
Tic observation[d]		6.50 (4.16)	7.68 (4.14)	7.09 (4.12)	0.44
Tic suppression[d]		6.77 (3.76)	5.60 (3.54)	6.19 (3.64)	0.39
PUTS total score		17.24 (5.95)	20.31 (7.44)	18.73 (6.79)	0.20
Neuropsychological functioning[b]	DCST	98.91 (13.38)	102.10 (11.66)	100.45 (12.48)	0.47
	Flanker test	98.23 (13.82)	98.81 (10.31)	98.51 (12.06)	0.89
	Motor dexterity	98.08 (11.64)	101.82 (14.69)[c]	99.83 (13.08)	0.43

***** *All two-tailed*
[a] *Equal variances assumed, with the exception of YGTSS motor scores*
[b] *Age-adjusted scaled scores*
[c] *N = 15 due to missing data*
[d] *N = 30 (CBIT = 15; Psych-Ed = 15) due to missing data*

Table 4: Categorical descriptive data at baseline with tests for group differences

		CBIT group (n = 17)	Psych-Ed group (n = 16)	Total sample (n = 33)	Fisher's Exact test
					p-value*
Gender	Male	13	12	25 (76%)	1.00
	Female	4	4	8 (24%)	
Handedness	Right-handed	3	3	6 (18%)	1.00
	Left-handed	14	13	27 (82%)	
Ethnicity[a]	White British	11	12	23 (70%)	0.71
	White - Other	4	3	7 (21%)	
	British - Indian	0	1	1 (3%)	
	British - Black	1	0	1 (3%)	
	Mixed / multiple ethnic	1	0	1 (3%)	
Tic disorder[b]	TS	15	15	30 (91%)	1.00
	CMTD	2	1	3 (9%)	

Any co-morbidity[c]	Yes	8	10	18 (55%)	0.49
	No	9	6	15 (45%)	
ADHD diagnosis[d]	Yes	2	5	7 (21%)	0.23
	No	15	11	26 (79%)	
ADHD-inattentive symptoms (SNAP-IV parent rating)	Clinical level	10	5	15 (45%)	0.17
	Non-clinical level	7	11	18 (55%)	
ADHD-hyperactive symptoms (SNAP-IV parent rating)	Clinical level	3	2	5 (15%)	1.00
	Non-clinical level	14	14	28 (85%)	
OCD diagnosis[d]	Yes	4	5	9 (27%)	0.71
	No	13	11	24 (73%)	
OCD symptom impairment (CHOCCI child self-report)[e]	Clinical level	5	8	13 (41%)	0.47
	Non-clinical level	11	8	19 (59%)	
OCD symptom impairment (CHOCCI parent self-report)	Clinical level	5	6	11 (33%)	0.72
	Non-clinical level	12	10	22 (67%)	
ASD diagnosis[d]	Yes	1	1	2 (6%)	1.00
	No	16	15	31 (94%)	
Other diagnoses[d,f]	Anxiety/panic	2	0	2 (6%)	-
	PTSD	1	0	1 (3%)	
	ODD	0	1	1 (3%)	
	Dyspraxia	1	1	2 (6%)	
	Dyscalculia	0	1	1 (3%)	
	Epilepsy	1	0	1 (3%)	
Currently taking medication? (within last 6 weeks)[d,g]	Yes	7	4	11 (33%)	0.47
	No	10	12	22 (67%)	
Had previous treatment?[d,h]	Yes	3	5	8 (24%)	0.44
	No	14	11	25 (76%)	
Having other current treatment?[d,i]	Yes	3	1	4 (12%)	0.60
	No	14	15	29 (88%)	
Month group began	September	10	6	16 (48%)	0.30
	November	7	10	17 (52%)	
Recruitment source	Retrospective	5	6	11 (33%)	0.72
	New referral	12	10	22 (67%)	

Notes. CMTD = Chronic Motor Tic Disorder; ODD = Oppositional Defiant Disorder
***** *All two-tailed*
[a] *Expected cell frequencies were <1 so data was pooled and analysed using a 2 x 2 contingency table of group type (CBIT / PE) vs ethnicity (White British vs other)*
[b] *As diagnosed in the TS clinic and verified by YGTSS ratings in the present study*
[c] *11 children had one co-morbidity, six had two, and one child had three co-morbidities*
[d] *Parent report at intake*
[e] *n = 32 (missing data)*
[f] *Numbers too few to conduct Fisher's exact test (expected cell frequencies <1)*

[g] *Ten children were prescribed non-stimulant medications; One child in the CBIT group was prescribed stimulant medication*
[h] *Six children were reported to have had one previous treatment; Two children had received two previous treatments. Of the previous treatments, four were CBT for other conditions such as anxiety (CBIT = 3; Psych-Ed = 1; duration 2 – 11 sessions, one unknown), two were the psycho-educational group (>2 years ago; CBIT = 0; Psych-Ed = 2), one was individual therapy sessions relating to tics (Psych-Ed = 1; 4 sessions), one was family therapy (Psych-Ed = 1; roughly 42 sessions), one was play therapy for anxiety (CBIT = 1; unknown duration), one was unspecified counselling (Psych-Ed = 1; 2 sessions) and one involved social stories for suspected Asperger's (Psych-Ed = 1, unknown duration)*
[i] *Current treatment: Four children were engaged in additional psychological therapies whist attending the groups (CBIT = 3; Psych-Ed = 1)*

Hypothesis testing: Primary analysis

In this study, ITT analysis (*n* = 33) is primarily reported. Should any significant changes be observed in the pattern of results when using an attenders-only analysis (*n* = 26) or following the removal of confounding cases (*see Method: Statistical Analysis*), these will be discussed.

Hypothesis 1: Children in the CBIT group will show significant pre- to post-treatment improvements in tic severity and neuropsychological functioning (response inhibition, cognitive flexibility and fine motor dexterity), compared with those in the psycho-educational (PE) group for whom little or no changes are expected

The findings of eight repeated-measures Analysis of Variance (ANOVA) tests, following a mixed 2 x 2 design as previously described, are outlined in Table 5. Findings relating to the PUTS measure (hypothesis 2) are also included to display the complete data set. The observed power for each variable tested within Table 5 is listed in Appendix 39.

Table 5: Descriptive data, significance tests and effect sizes for hypotheses one and two

		CBIT group (*n* = 17)		Psych-Ed group (*n* = 16)		Sig. main effect of group type?[a]	Sig. main effect of time?[a]	Sig. interaction?[a]	Total sample (*n* = 33)	
		Pre	*Post*	*Pre*	*Post*	*p-value (effect size)*	*p-value (effect size)*	*p-value (effect size)*	*Pre*	*Post*
		M (SD), 95% CI	*M (SD), 95% CI*	*M (SD), 95% CI*	*M (SD), 95% CI*				*M (SD), 95% CI*	*M (SD), 95% CI*
YGTSS	*Motor*	17.65 (4.74), 15.67 - 19.63	15.12 (4.30), 13.40 - 16.84	16.31 (3.03), 14.27 - 18.36	15.88 (2.28), 14.11 - 17.65	0.82	0.001** (η_p^2=0.31, large)	0.01** (η_p^2=0.18, medium)	16.98 (0.70), 15.56 - 18.40	15.50 (0.61), 14.26 - 16.73
	Phonic	12.71 (6.99), 9.49 - 15.92	12.71 (5.61), 9.88 - 15.53	12.63 (5.93), 9.31 - 15.94	11.13 (5.82), 8.21 - 14.04	0.68	0.37	0.37	12.67 (6.40), 10.36 - 14.97	11.94 (5.68), 9.89 - 13.94
	Total (with impairment)	54.47 (16.43), 46.80 - 62.15	44.88 (16.78), 37.59 - 52.17	50.19 (14.48), 42.28 - 58.10	47.00 (12.19), 39.48 - 54.52	0.82	0.01** (η_p^2=0.20, medium)	0.17	52.39 (15.43), 46.82 - 57.84	45.91 (14.55), 40.71 - 51.18
	Tic observation	6.50 (4.16), 4.39 - 8.62	4.92 (3.83), 3.23 - 6.62	7.18 (3.80), 5.00 - 9.37	5.60 (2.35), 3.85 - 7.36	0.58	0.01** (η_p^2=0.24, medium)	1.00	6.83 (3.94), 5.32 - 8.37	5.25 (3.16), 4.04 - 6.49
	Tic suppression	6.77 (3.76), 4.83 - 8.71	5.17 (3.37), 3.57 - 6.78	5.37 (3.55), 3.36 - 7.38	4.30 (2.60), 2.64 - 5.96	0.34	0.01** (η_p^2=0.25, medium)	0.56	6.10 (3.67), 4.68 - 7.47	4.75 (3.00), 3.58 - 5.89
PUTS		17.24 (5.95), 13.92 - 20.56	18.41 (6.65), 14.89 - 21.94	20.31 (7.44), 16.89 - 23.74	21.69 (7.60), 18.05 - 25.32	0.17	0.11	0.90	18.73 (6.79), 16.39 - 21.16	20.00 (7.21), 17.52 - 22.58
Neuropsych. functioning	*DCST*	98.23 (13.51), 91.78 - 104.67	98.26 (12.06), 92.12 - 104.39	102.10 (11.66), 95.65 - 108.54	101.29 (11.98), 95.15 - 107.43	0.36	0.87	0.86	100.16 (12.57), 95.61 - 104.72	99.77 (11.92), 95.43 - 104.11
	Flanker test	97.51 (13.95), 91.25 - 103.77	100.05 (13.77), 93.88 - 106.21	98.81 (10.31), 92.55 - 105.07	100.70 (10.10), 94.53 - 106.87	0.80	0.29[b]	0.88	98.16 (12.08), 93.73 - 102.59	100.37 (11.89), 96.01 - 104.73
	Motor dexterity	98.08 (11.64), 91.56 - 104.59	98.36 (14.48), 91.94 - 104.78	101.82 (14.69), 94.89 - 108.76	100.15 (10.97), 93.32 - 106.99	0.50	0.75	0.66	99.83 (13.08), 95.19 - 104.71	99.20 (12.78), 94.57 - 103.94

Notes. CBIT = Comprehensive Behavioural Intervention for Tics group; Psych-Ed = Psycho-educational group; Sig. = Significant; Neuropsyc. = Neuropsychological; η_p^2 = Partial eta-squared

*** Significant result that survives Bonferroni correction (0.05/8 = 0.01). The PUTS measure is not involved in the primary analysis so is not included in Bonferroni correction calculations.*

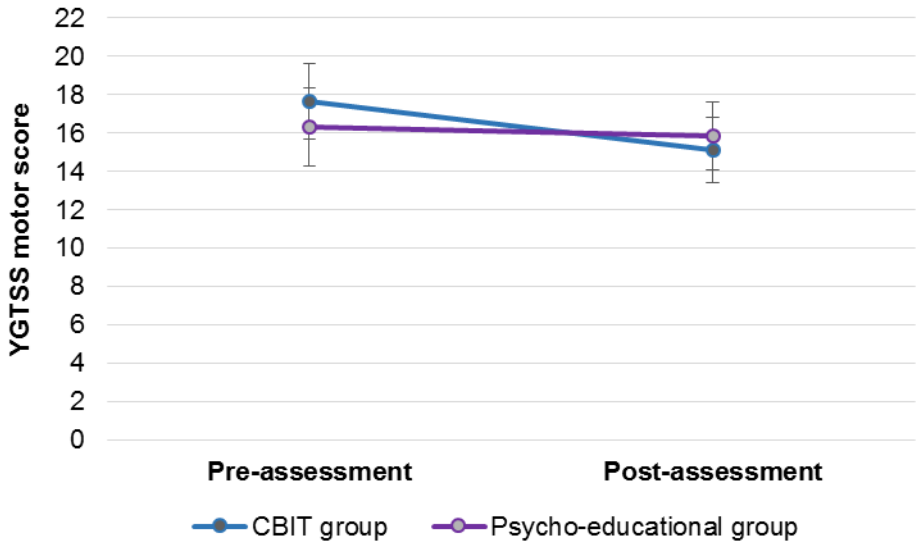
[a] Repeated-Measures Analysis of Variance (ANOVA)

[b] Significant finding when medication change cases removed, n = 5 (p = 0.02; does not survive Bonferroni correction; η_p^2 = 0.20, medium effect size)

Tic outcomes: YGTSS

On the YGTSS motor scale there was a significant main effect of time, in which children in both groups demonstrated reductions in motor tics (14.3% versus 2.6% in CBIT and PE groups respectively) from pre- to post-assessment ($F(1,31) = 13.87$, $p = 0.001$, $\eta_p^2=0.31$, large effect size). Although the main effect of group type (CBIT versus PE) was non-significant ($F(1,31) = 0.05$, $p = 0.82$), there was a significant group type by time interaction ($F(1,31) = 6.90$, $p = 0.01$, $\eta_p^2=0.18$, medium effect size). Both findings survived Bonferroni correction ($p \leq 0.01$). This indicated that over the course of treatment, children in the CBIT group improved significantly more in terms of motor tic severity compared with those in the PE group (see Figure 3). Although ANOVA tests are fairly robust to the violation of assumptions, these findings must be interpreted with some caution. A significant Levene's test (pre-assessment: $F(1,31) = 6.65$, $p = 0.02$; post-assessment: $F(1,31) = 10.17$, $p = 0.003$) indicated unequal variances. Sphericity issues were not applicable due to the 2 x 2 design of the repeated-measures ANOVA. There was no clear skew direction and transformations (squareroot and logarithmic) were unsuccessful.

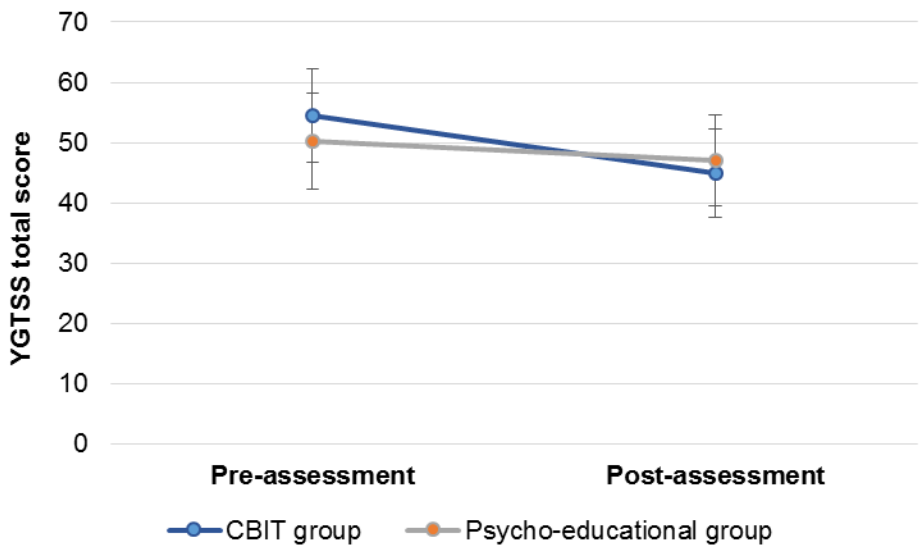
Figure 3: Significant interaction (group type x time) and main effect of time on the YGTSS motor subscale



On the YGTSS phonic scale, children in the CBIT group experienced no change in phonic tics from pre-to post-assessment, whereas those in the PE group experienced an 11.9% reduction. There was no significant main effect of time (F(1,31) = 0.82, *p* = 0.37) or group type (F(1,31) = 0.18, *p* = 0.68). There was no significant group type by time interaction (F(1,31) = 0.82, *p* = 0.37).

On YGTSS total scores there was a significant main effect of time, reflecting significant reductions in tics for both groups (17.6% for CBIT versus 6.4% for PE groups) from pre- to post-assessment (F(1,31) = 7.83, *p* = 0.01, η_p^2=0.20, medium effect size). This finding survived Bonferroni correction (p≤0.01) and is displayed in Figure 4. The differences in improvement between the two groups was non-significant, with no group type by time interactions (F(1,31) = 1.97, *p* = 0.17) or main effects of group type (F(1,31) = 0.05, *p* = 0.82).

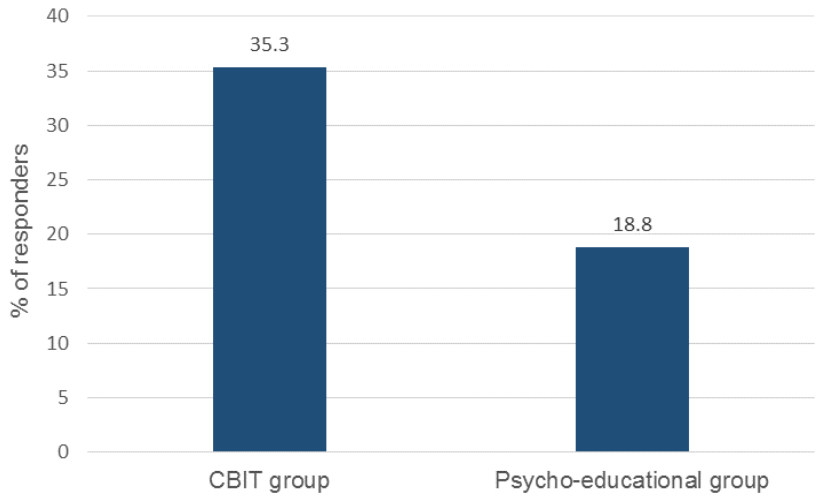
Figure 4: Main effect of time with YGTSS total scores



Jeon and colleagues (2013) suggest that a 25% reduction in tic severity, as measured by the combination of scores on the motor and phonic (referred to in this report as "combined tic severity score") subscales, represents clinically meaningful change. According to this criterion on an individual basis, 35.3% (*n* = 6; range of tic reduction = 25.8% to 47.1%) of children in the CBIT group would be classified as treatment

"responders", in comparison to 18.8% "responders" (*n* = 3; range of tic reduction = 25.64% to 29.63%) in the PE group. This is displayed in Figure 5.

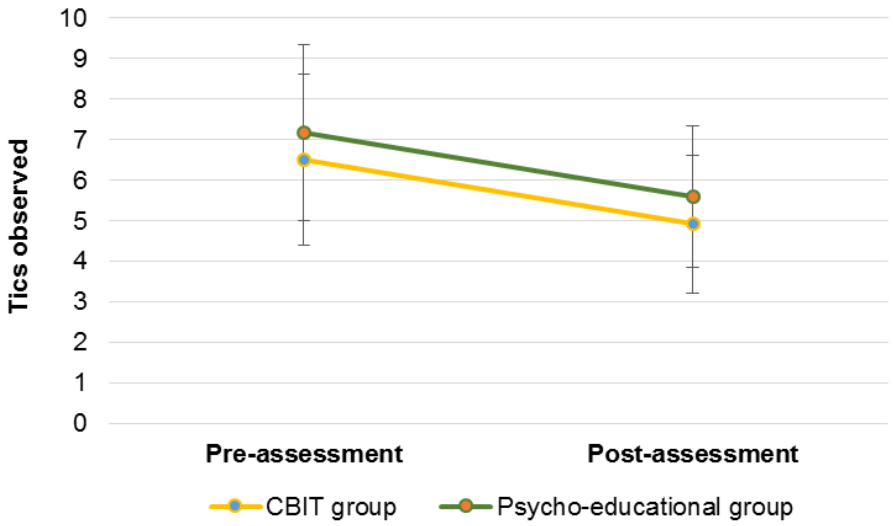
Figure 5: Treatment responders in CBIT and PE groups using YGTSS total scores



Tic outcomes: Tic observation and suppression

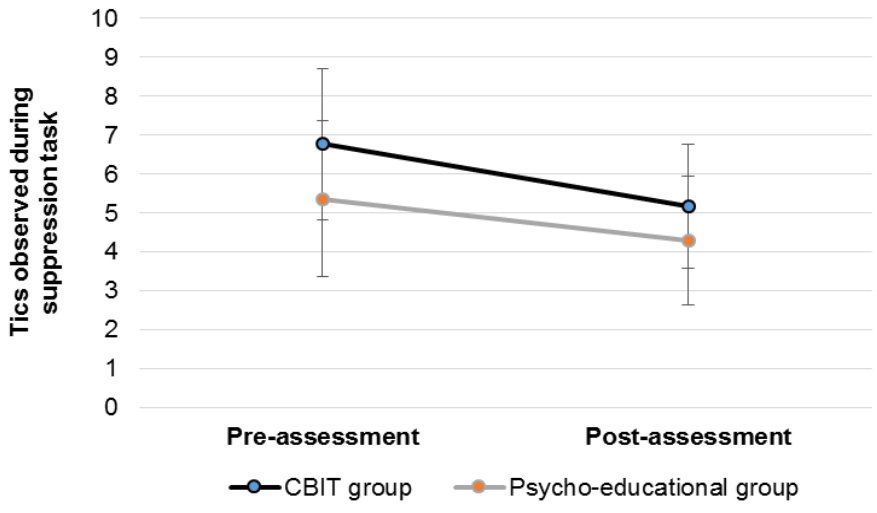
On the direct tic observation measure, children in both groups experienced significant reductions in tics from pre- to post-assessment (24.3% for CBIT versus 22.0% for psycho-educational groups), as evidenced by a significant main effect of time ($F(1,27) = 8.34$, $p = 0.01$, $\eta_p^2=0.24$, medium effect size). This survived Bonferroni correction ($p \leq 0.01$) and is displayed in Figure 6. Although children in the CBIT group appeared to improve more, findings were non-significant for main effect of group type ($F(1,27) = 0.31$, $p = 0.58$) and interaction of group type by time ($F(1,27) = 0.00$, $p = 1.00$).

Figure 6: Main effect of time with direct tics observed



On the tic suppression task, children in both groups experienced a significant reduction in observed tics from pre- to post-assessment (23.6% versus 19.9% for CBIT and psycho-educational groups respectively), as evidenced by a significant main effect of time ($F(1,27) = 8.87$, $p = 0.01$, $\eta_p^2=0.25$, medium effect size). This finding survived Bonferroni correction ($p\leq0.01$) and is displayed in Figure 7. Children in the CBIT group appeared to improve more, however findings were non-significant for main effect of group type ($F(1,27) = 0.95$, $p = 0.34$) and interaction of group type by time ($F(1,27) = 0.35$, $p = 0.56$).

Figure 7: Main effect of time with tics observed during the suppression task



Adverse effects of groups on tics

Feedback from satisfaction questionnaires, which were routinely given out in clinical practice, indicated that parents were equally satisfied with both types of groups (Ince, Shafran & Murphy, in preparation). Data on child satisfaction was unfortunately not available in the present study. While feedback was positive, individual pre-post change scores on the YGTSS[13] provided a further objective evaluation of any adverse effects of the groups on children's tics. An evaluation of adverse effects is recommended by the CONSORT guidelines for non-pharmacological randomised controlled trials (Boutron et al., 2008). YGTSS scores were used as they took into account the entire week preceding assessment, which is important given the characteristic fluctuating nature of tics.

On the YGTSS motor subscale, only one child (6%) in the CBIT group experienced a slight worsening in motor tics, compared with five children (31%) in the PE group[14]. On the YGTSS phonic subscale, six children (35%) in the CBIT group experienced a worsening in phonic tics[15], compared with two children (13%) in the PE group[16]. On the YGTSS total scores, two children (12%) in the CBIT group experienced a worsening in tics[17], compared with five children (31%) in the PE group.

Overall, the large majority of children in both groups did not experience any deterioration in tics from pre- to post-assessment. Those in the CBIT group

[13] Calculated through time two minus time one change scores for each child, so that negative values indicate a deterioration.

[14] Two cases may have been affected by confounding variables (medication change and a significant life event).

[15] Data from two children who experienced the largest deteriorations is likely to have been affected by confounding variables (medication change and a significant life event).

[16] Data from one child is likely to have been affected by a confounding variable (medication change).

[17] One of which was marginal and the other confounded by a change in medication.

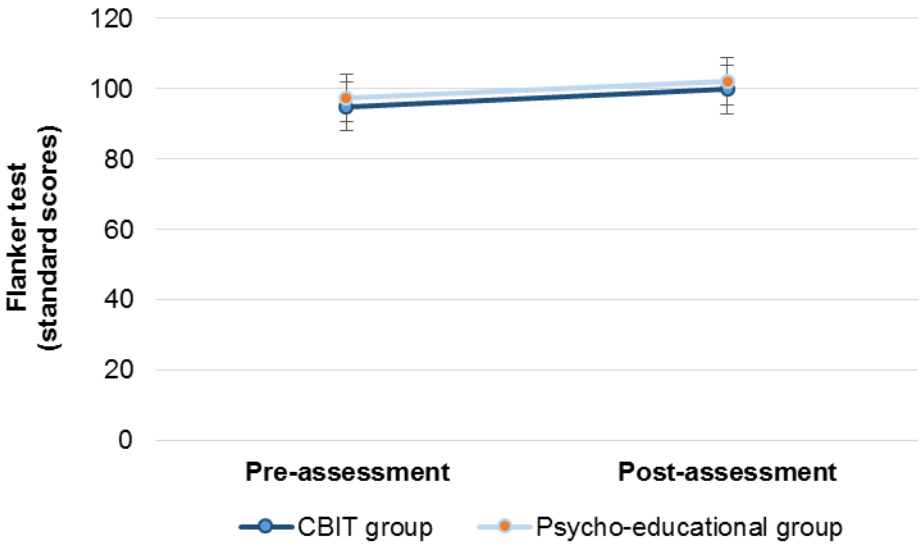
experienced the fewest adverse effects in comparison to the PE group, with the exception of the YGTSS phonic tics subscale.

Neuropsychological functioning outcomes

On the DCST measure of attention, cognitive flexibility and response inhibition, children in the CBIT groups experienced no change in standard scores achieved from pre-to post-assessment. This was in comparison with a 0.8% marginal improvement in the PE groups. There were no main effects of time ($F(1,30) = 0.03$, $p = 0.87$), group type ($F(1,30) = 0.87$, $p = 0.36$) or interactions of group type by time ($F(1,30) = 0.03$, $p = 0.86$).

On the Flanker test of inhibitory control and attention, children in the CBIT groups experienced a 2.6% improvement in standard scores achieved from pre-to post-assessment. Children in the PE group demonstrated a 1.9% improvement. There were no significant main effects of group type ($F(1,30) = 0.07$, $p = 0.80$) or interactions of group type by time ($F(1,30) = 0.03$, $p = 0.88$). Both groups displayed small improvements, although main effect of time was non-significant ($F(1,30) = 1.18$, $p = 0.29$). When cases of children who had experienced a change in medication were removed, the main effect of time became significant ($F(1,25) = 6.35$, $p = 0.02$, $\eta_p^2 = 0.20$, medium-to-large effect size), however did not reach Bonferroni-adjusted significance levels ($p \leq 0.01$). This finding is displayed in Figure 8.

Figure 8: Main effect of time on Flanker test (standard scores)



Note: Medication change cases (n = 5) removed

On the pegtest children in the CBIT group experienced a 0.3% improvement in standard scores achieved from pre-to post-assessment, compared with 1.6% in the PE groups. There were no significant main effects of time (F(1,30) = 0.10, *p* = 0.75), group type (F(1,30) = 0.46, *p* = 0.50) or interactions of group type by time (F(1,30) = 0.20, *p* = 0.66).

Hypothesis testing: Secondary analyses

Hypothesis 2: Children in the CBIT group will show a significant increase in PU awareness from pre- to post-intervention, compared with those in the PE group for whom little or no changes are expected

Children in both the CBIT and PE groups experienced a 6.8% increase in PU awareness from pre- to post-assessment, however there was no main effect of time (F(1,31) = 2.73, *p* = 0.11). There was also no main effect of group type (F(1,31) = 1.93, *p* = 0.17) or group type by time interactions (F(1,31) = 0.02, *p* = 0.90).

Hypothesis 3a: At baseline, higher levels of neuropsychological functioning (response inhibition, cognitive flexibility, fine motor dexterity, motor-based processing speed) will be correlated with better tic suppression scores and lower tic severity

An ITT analysis[18] using Pearson's correlations was conducted with selected variables at baseline, for all children irrespective of group. YGTSS (motor, phonic and total) scores, as well as tic observation and suppression tasks, measured tic severity. All correlations are displayed in Table 6.

Baseline scores on the Flanker test were significantly and negatively associated, as expected, with tic severity as measured by tics counted in observation (r = -0.48, large effect size, p = 0.01) and suppression (r = -0.38, medium effect size, p = 0.04) conditions. Only the former met the Bonferroni-adjusted significance level (p≤0.01) and is displayed in Figure 9. The findings indicate that poor response inhibition at baseline was associated with worse baseline tic severity in terms of tics observed and suppression ability.

Baseline scores on the DCST test were also significantly and negatively associated, as expected, with tic severity as measured by suppression ability (r = -0.40 (medium effect size), p = 0.03). This did not meet the Bonferroni-corrected significance level (p≤0.01).

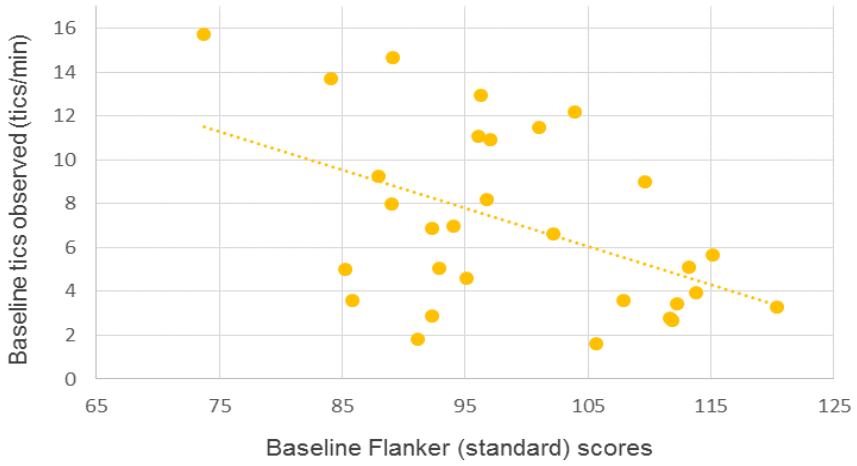
[18] Secondary analyses (i.e. attenders-only and removal of potentially confounding cases) were not considered appropriate due to measures being at baseline. The only analysis that was still carried out was removing a case on stimulant medication (n = 1), which did not indicate a different pattern of results and thus is not discussed.

Table 6: Correlations at baseline between neuropsychological functioning, tic severity and suppression scores (n = 33)

		T1 DCST	T1 Flanker	T1 pegboard	T1 WISC-IV coding	T1 direct tic obs (tics/min)	T1 tic suppression (tics/min)	T1 YGTSS motor	T1 YGTSS phonic	T1 YGTSS total
T1 DCST	Pearson correlation					-0.20	-0.40	-0.19	-0.05	-0.22
	Sig. (2-tailed)					0.29	0.03*	0.30	0.78	0.21
T1 Flanker	Pearson correlation					-0.48	-0.38	-0.17	0.15	0.07
	Sig. (2-tailed)					0.01**	0.04*	0.36	0.42	0.69
T1 pegboard	Pearson correlation					0.22	0.24	0.04	0.19	0.11
	Sig. (2-tailed)					0.25	0.21	0.82	0.29	0.56
T1 WISC-IV coding	Pearson correlation					0.32	0.12	0.03	0.05	0.00
	Sig. (2-tailed)					0.09	0.54	0.85	0.80	0.99
T1 direct tic obs (tics/min)	Pearson correlation	-0.20	-0.48	0.22	0.32					
	Sig. (2-tailed)	0.29	0.01**	0.25	0.09					
T1 tic suppression (tics/min)	Pearson correlation	-0.40	-0.38	0.24	0.12					
	Sig. (2-tailed)	0.03*	0.04*	0.21	0.54					
T1 YGTSS motor	Pearson correlation	-0.19	-0.17	0.04	0.03					
	Sig. (2-tailed)	0.30	0.36	0.82	0.85					
T1 YGTSS phonic	Pearson correlation	-0.05	0.15	0.19	0.05					
	Sig. (2-tailed)	0.78	0.42	0.29	0.80					
T1 YGTSS total	Pearson correlation	-0.22	0.07	0.11	0.00					
	Sig. (2-tailed)	0.21	0.69	0.56	0.99					

** Significant at p ≤ 0.05*
*** Significant at p ≤ 0.01 (Bonferroni-adjusted)*
Note. T1 = Time 1 (pre-assessment); DCST = Dimensional Card Sort Test; YGTSS = Yale Global Tic Severity Scale; Obs = Observation

Figure 9: The association between baseline scores on the Flanker test and baseline tics observed



Hypothesis 3b: Pre- to post-treatment changes on measures of neuropsychological functioning (response inhibition, cognitive flexibility and fine motor dexterity) will be positively correlated with changes in tic severity and suppression ability (CBIT group only)

The eight variables of interest were YGTSS (motor, phonic and total), tics observed, suppression ability and neuropsychological functioning (response inhibition, cognitive flexibility and fine motor dexterity) change scores[19], from pre- to post-assessment. Pearson partial correlations were used, controlling for the baseline scores of all eight variables. Hypothesis 3b was intended to include participants from the CBIT group only. However, as improvements were observed in both groups on both types of variables (tic severity and neuropsychological), it was deemed appropriate to conduct an analysis using the whole sample (n = 33).

Whole sample analysis

[19] Changes in neuropsychological functioning were calculated as time two minus time one, so that positive scores indicated changes in the expected direction. Changes on the YGTSS, tic observations and suppression ability were calculated as time one minus time two, again so that positive scores indicated changes in the expected direction.

An ITT analysis with children in both groups (n = 33) indicated a significant positive association between YGTSS motor and DCST change scores only (r = 0.50, large effect size, p = 0.03). All correlations are displayed in Table 7. This finding was no longer significant following an attenders-only analysis (r = 0.32, p = 0.24). Upon the removal of cases in the ITT analysis who had experienced significant life events, there was a significant positive correlation between change scores on the YGTSS total and fine motor dexterity measures (r = 0.55, large effect size, p = 0.02). When cases receiving additional current psychological input were removed, the significant association between DCST and YGTSS motor change scores was no longer significant (r = 0.46, p = 0.08). This finding was similar following the removal of cases whose post-assessment had been delayed (r = 0.38, p = 0.13). When medication change cases were removed, the significant association between the DCST and YGTSS motor change scores were no longer significant (r = 0.47, p = 0.09). However, a new significant positive correlation was indicated between the DCST and YGTSS total change scores (r = 0.67, p = 0.01, large effect size). This was the only finding to meet the Bonferroni-adjusted significance levels ($p \leq 0.01$) and is displayed in Figure 10. Upon inspection of Figure 10, it can be noted that eight cases demonstrate unexpected associations. Two indicate that a deterioration on the DCST was associated with an improvement in tic severity. Six indicate that an improvement on the DCST was associated with a deterioration in tics. However, the overall sample demonstrated positive associations in the expected direction.

Table 7: Correlations between neuropsychological functioning, tic severity and suppression change scores (pre- to post-assessment)

		YGTSS motor changes	YGTSS phonic changes	YGTSS total changes	Tic obs changes (tics/min)	Tic suppression changes (tics/min)	DCST changes	Flanker changes	Pegboard changes
YGTSS motor changes	Pearson correlation						0.50	0.28	0.15
	Sig. (2-tailed)						0.03*[a]	0.25	0.53
YGTSS phonic changes	Pearson correlation						0.20	0.04	-0.08
	Sig. (2-tailed)						0.41	0.89	0.76
YGTSS total changes	Pearson correlation						0.34[c]	-0.07	0.41[b]
	Sig. (2-tailed)						0.15	0.79	0.08
Tic obs changes (tics/min)	Pearson correlation						0.13	-0.29	-0.07
	Sig. (2-tailed)						0.60	0.23	0.76
Tic suppression changes (tics/min)	Pearson correlation						0.44	-0.13	0.17
	Sig. (2-tailed)						0.06	0.59	0.49
DCST changes	Pearson correlation	0.50	0.20	0.34[c]	0.13	0.44			
	Sig. (2-tailed)	0.03*[a]	0.41	0.15	0.60	0.06			
Flanker changes	Pearson correlation	0.28	0.04	-0.07	-0.29	-0.13			
	Sig. (2-tailed)	0.25	0.89	0.79	0.23	0.59			
Pegboard changes	Pearson correlation	0.15	-0.08	0.41[b]	-0.07	0.17			
	Sig. (2-tailed)	0.53	0.76	0.08	0.76	0.49			

Note. Pearson partial correlations were conducted, controlling for baseline tic severity and suppression scores. YGTSS = Yale Global Tic Severity Scale; Obs = Observation; DCST = Dimensional Card Sort Test.

** Significant at p ≤ 0.05*

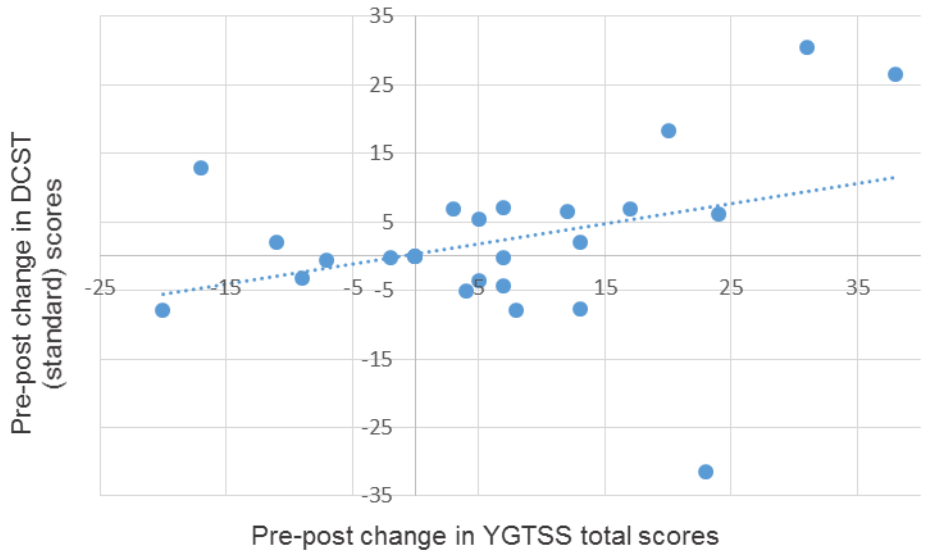
*** Significant at p ≤ 0.01 (Bonferroni-adjusted)*

[a] Did not survive attenders-only analysis (n=26) and became non-significant upon removal of cases receiving additional current psychological input (n=4), those who had experienced a change in medication (n=5) and children whose post-assessment had been significantly delayed (n=2).

[b] Significant when cases removed who had experienced significant life events (n=3).

[c] Significant when medication change cases removed (n=5)

Figure 10: ITT analysis of the association between DCST and YGTSS total pre- to post-treatment change scores, following the removal of medication change cases (total n for analysis = 28)



Hypothesis 4: Higher baseline neuropsychological functioning (response inhibition, cognitive flexibility, fine motor dexterity, motor-based processing speed) will correlate with, and predict, greater pre-post improvements in tic severity and suppression ability (CBIT group only)

The variables of interest for this analysis were YGTSS (motor, phonic and total) change scores and changes in tics per minute (observation and suppression) from pre- to post-assessment[20]. Including measures of neuropsychological functioning at baseline (DCST, Flanker, pegtest and WISC coding), a total of nine variables were of interest. Pearson partial correlations were used, controlling for the baseline (pre-assessment) scores of all five tic severity and suppression measures.

An ITT analysis with children in the CBIT group only (*n* = 17) demonstrated a significant positive association between baseline fine motor dexterity and YGTSS motor change scores only (*r* = 0.82, large effect size, *p* = 0.004). This survived

[20] Changes in tic severity and suppression ability were calculated as time one minus time two, again so that positive scores indicated changes in the expected direction.

Bonferroni correction and is displayed in Figure 11. All correlations are displayed in Table 8.

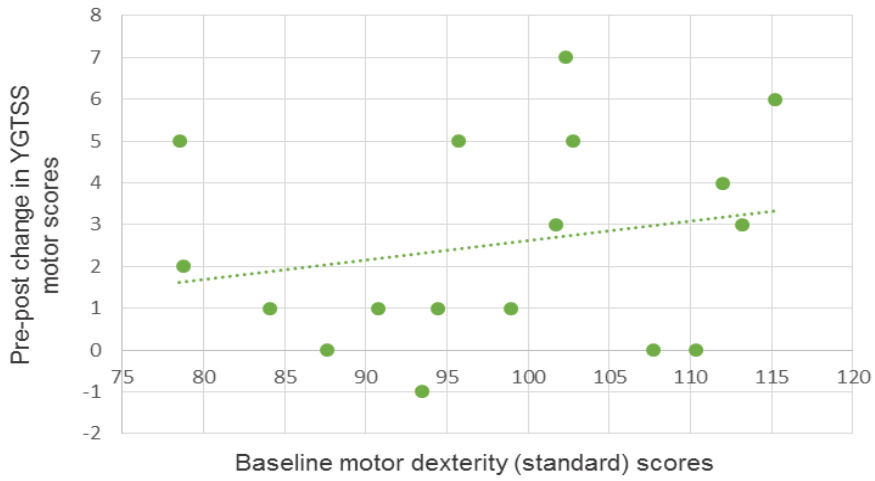
Table 8: Correlations of baseline neuropsychological functioning with tic severity and suppression change scores (pre- to post-assessment)

		Baseline DCST	Baseline Flanker	Baseline Pegboard	YGTSS motor changes	YGTSS phonic changes	YGTSS total changes	Tic obs changes (tics/min)	Tic suppression changes (tics/min)
Baseline DCST	Pearson correlation				-0.03	-0.51	-0.16	-0.12	0.00
	Sig. (2-tailed)				0.94	0.14	0.66	0.75	0.99
Baseline Flanker	Pearson correlation				0.39	-0.42	-0.23	-0.31	-0.25
	Sig. (2-tailed)				0.27	0.23	0.52	0.38	0.49
Baseline Pegboard	Pearson correlation				0.82**	-0.08	0.61	-0.09	0.25
	Sig. (2-tailed)				0.004	0.82	0.06	0.81	0.50
YGTSS motor changes	Pearson correlation	-0.03	0.39	0.82**					
	Sig. (2-tailed)	0.94	0.27	0.004					
YGTSS phonic changes	Pearson correlation	-0.51	-0.42	-0.08					
	Sig. (2-tailed)	0.14	0.23	0.82					
YGTSS total changes	Pearson correlation	-0.16	-0.23	0.61					
	Sig. (2-tailed)	0.66	0.52	0.06					
Tic obs changes (tics/min)	Pearson correlation	-0.12	-0.31	-0.09					
	Sig. (2-tailed)	0.75	0.38	0.81					
Tic suppression changes (tics/min)	Pearson correlation	0.00	-0.25	0.25					
	Sig. (2-tailed)	0.99	0.49	0.50					

** Significant at p ≤ 0.05*
*** Significant at p ≤ 0.01 (Bonferroni-adjusted)*
Note: Pearson partial correlations were conducted, controlling for baseline tic severity and suppression scores. DCST = Dimensional Card Sort Test; YGTSS = Yale Global Tic Severity Scale; Obs = Observation

This finding was no longer significant when medication change cases (*n* = 2) were removed (*r* = 0.73, *p* = 0.06), however did survive an attenders-only (n = 15) analysis (*r* = 0.82, *p* = 0.004).

Figure 11: The association between baseline fine motor dexterity ability and pre- to post-treatment change scores on the YGTSS motor subscale (CBIT group only)



Hypothesis 5: Lower baseline ADHD symptomatology will correlate with, and predict, greater pre-post improvements in tic severity and suppression ability (CBIT group only)

The variables of interest for this were pre-post change scores on the YGTSS (motor, phonic and total) and tics observed or suppressed[21]. Baseline ADHD symptomatology was measured by the SNAP-IV, examining inattentive and hyperactive/impulsive subscales separately. Thus, in total there were seven variables of interest. Pearson partial correlations were used, controlling for the baseline (pre-assessment) scores of all five tic severity variables representing change from pre- to post-assessment.

An ITT analysis with children in the CBIT group only (*n* = 17) did not demonstrate any significant associations, as displayed in Table 9. This was also the case using an attenders-only (*n* = 15) analysis.

[21] Changes in tic severity and suppression ability were calculated as time one minus time two, again so that positive scores indicated changes in the expected direction.

Discussion

The primary aim of this exploratory pilot study was to evaluate the feasibility and preliminary effectiveness of newly-manualised group-based CBIT and psycho-educational (PE) interventions, for children aged 9 to 13 years old with TS and chronic tic disorders. The main findings of all analyses will be discussed, followed by an evaluation of the strengths and limitations of this study, conclusions and ideas for future research.

Key Findings

Feasibility

Participant uptake to this study was good (30%), with more children being recruited as new referrals than those invited retrospectively. Low drop-out rates (9%) and regular attendance in both groups suggested good feasibility and acceptability of these interventions. Attendance in the CBIT group was better than the PE group, with 88% and 69% of children respectively attending at least five or more sessions ("attenders").

Table 9: Correlations of baseline ADHD symptomatology with tic severity and suppression change scores (pre- to post-assessment)

		Baseline ADHD-Inattention score	Baseline ADHD-Hyperactivity/ Impulsivity score	YGTSS motor changes	YGTSS phonic changes	YGTSS total changes	Tic obs changes (tics/min)	Tic suppression changes (tics/min)
Baseline ADHD-Inattention score	Pearson correlation			-0.14	-0.54	-0.03	-0.27	-0.26
	Sig. (2-tailed)			0.79	0.27	0.96	0.60	0.62
Baseline ADHD-Hyperactivity/ Impulsivity score	Pearson correlation			-0.24	-0.32	0.08	-0.02	0.01
	Sig. (2-tailed)			0.64	0.54	0.88	0.97	0.98
YGTSS motor changes	Pearson correlation	-0.14	-0.24					
	Sig. (2-tailed)	0.79	0.64					
YGTSS phonic changes	Pearson correlation	-0.54	-0.32					
	Sig. (2-tailed)	0.27	0.54					
YGTSS total changes	Pearson correlation	-0.03	0.08					
	Sig. (2-tailed)	0.96	0.88					
Tic obs changes (tics/min)	Pearson correlation	-0.27	-0.02					
	Sig. (2-tailed)	0.60	0.97					
Tic suppression changes (tics/min)	Pearson correlation	-0.26	0.01					
	Sig. (2-tailed)	0.62	0.98					

Note: Pearson partial correlations were conducted, controlling for baseline tic severity and suppression scores. YGTSS = Yale Global Tic Severity Scale; Obs = Observation

Overall this study provided supportive evidence that CBIT and PE groups can both lead to significant improvements in tic severity for children with TS, as measured by indirect (YGTSS) and direct (tic observation and suppression) measures. This was unexpected, as it had been assumed that only the CBIT group would lead to improvements, considering the PE group did not focus on reducing or managing tics. All tic outcomes in the primary analysis met Bonferroni-adjusted significance levels ($p \leq 0.01$), which controlled for the inflated Type I error rate resulting from multiple comparisons. The medium to large effect sizes observed indicated clinically meaningful change and were comparable to treatment outcomes in individually-delivered HRT and CBIT treatments as well as antipsychotic medication trials (McGuire et al., 2014).

The exception to this finding was on the YGTSS motor tics subscale, in which an additional significant interaction indicated that children in the CBIT group improved significantly more in terms of motor tic severity (14.3% reduction) than those in the PE group (2.6% reduction). This may be explained by the fact that in the CBIT groups, 73% of the tics that children chose to work on were motor tics and only 27% were phonic tics. Thus, the full benefits of CBIT treatment may not have been observed with phonic tics, which most likely explains the non-significant result for this subscale. This is also likely to explain why children in the CBIT groups demonstrated low rates of adverse effects on the YGTSS motor and total tic severity subscales, compared with higher rates on the phonic subscales.

The significant interaction indicating CBIT to be a superior treatment to PE in reducing tics supports previous findings (Piacentini et al., 2010; Wilhelm et al., 2012). The tic reductions were less than those previously reported in individually-administered HRT and CBIT treatments for children (Verdellen et al., 2011a: 30 – 100% reductions; Piacentini et al., 2010: YGTSS motor score reductions of 27% and 14% for CBIT and PE interventions respectively). This indicates that the effect of

group interventions may be diluted in comparison to individual treatments. However, the medium effect size observed ($\eta_p^2 = 0.18$) is comparable to individually-delivered CBIT treatment for children when compared with psycho-education and supportive therapy (Piacentini et al., 2010) and suggests meaningful clinical change. Additionally, 35.3% of children in the CBIT group were classified as “treatment responders”, in comparison to only 18.8% in the PE groups. As this figure takes into account phonic tics, which did not demonstrate reductions in this study, these responder rates are likely to be a conservative estimate.

The finding that children in both groups improved significantly in terms of tic severity may be explained by several possible reasons. One explanation could be that one or more common factors between the two groups led to improvements in tic severity. For example, the parent groups or peer support and social learning factors. Both groups received brief psycho-education about tics and tic treatments, which families may have looked into further in their own time. It may have been useful to administer a measure of knowledge regarding HRT-based ideas and techniques at the end of the study, to gauge whether the two groups differed significantly in their knowledge and implementation of these techniques in the home environment. Expectancy effects may have influenced outcomes, with all participants receiving treatment at a national specialist centre with frequent therapist contact. Lastly, the findings could be an illustration of the Hawthorn effect (Roethlisberger & Dickson, 1939), which suggests that behaviours can change simply as a result of being observed. This is less likely for the YGTSS data which was retrospective.

It is also possible that the improvements in tic severity were achieved through different pathways or mechanisms of change. For example, children in the CBIT groups may have experienced reductions in tic severity due to the specific techniques taught. In comparison, those in the PE groups may have experienced tic severity improvements indirectly as a result of factors such as increased coping ability or

decreased stress and anxiety (particularly in relation to managing co-morbid difficulties). This is supported by evidence from Conelea and Woods (2008) indicating that factors such as stress and anxiety can influence the severity and expression of tics.

These findings are tentative and interpreted with caution, particularly due to the lack of assessment of blinding success on tic observation measures. Inter-rater reliability for tic observation data and the YGTSS were also of a questionable range, although this is partially controlled within the repeated-measures design by the same researchers and raters scoring the same children.

Adverse short-term effects of group treatment on tics

While the majority of children in both groups experienced significant improvements in tic severity, some demonstrated a worsening of tics (CBIT group: 6 – 35%; PE: 13 – 31%). These rates are higher than those observed by Piacentini and colleagues (2010) using individually-delivered CBIT, who reported rates of 1.6% and 6.2% in CBIT and PE groups respectively. However, Piacentini and colleagues only counted self-reported significant worsening of tics above and beyond normal fluctuations, as opposed to the present study which used a standardised measure to count even slight deteriorations.

Premonitory urge

One key aim of CBIT is to increase PU awareness in order to successfully “catch” and suppress tics. In this study, PU awareness was found to increase in both groups from pre- to post-assessment, although this was non-significant. This was unexpected; however it is possible that children in the psycho-educational groups experienced an increase in PU awareness simply by engaging in discussions about tics and the PU (which was discussed during the first session involving basic psycho-education in both groups).

Neuropsychological outcomes

The Flanker test of inhibitory control and attention was the only measure of neuropsychological functioning to demonstrate significant improvements from pre- to post-treatment. This was unexpectedly found in both the CBIT and PE groups, with mean standard scores increasing by 2.54 and 1.89 points respectively. While it did not quite reach the Bonferroni-adjusted significance level ($p \leq 0.01$), a medium effect size ($\eta_p^2 = 0.20$) suggested clinically meaningful change. This finding was dependent upon the removal of data from children ($n = 5$) who had changes in medication during the study.

This finding echoes previous research by Deckersbach and colleagues (2014), in which behavioural data (faster reaction times) on a visuospatial priming task similar to the task in the present study, indicated a main effect of time for CBIT attendees with TS and wait list controls. It was suggested that this could be the result of practice effects. This could also be the case with the Flanker test in the present study, however is unlikely due to the longer time period between testing (8 – 16 weeks versus 10 weeks in the Deckersbach et al. trial). It is unfortunate that practice effect data is not available from the NIH toolbox at present and there are no guidelines regarding recommended test-retest periods. It would have been helpful to be able to compare findings from the present study with a waitlist control group. It is encouraging that practice effects did not seem apparent on measures of cognitive flexibility (DCST) and fine motor skill (pegtest).

An alternative explanation could be that improvement in the inhibitory processes of children in both groups would be expected, given that both groups demonstrated significant improvements in tic severity. Changes in tic severity are likely to be associated with changes in neural pathways and brain structures responsible for movement and inhibitory control, such as the striatum. These changes should subsequently influence performance on behavioural measures of response inhibition

such as the Flanker test. Thus this study provides very tentative support for learning-induced neuroplasticity as measured using a neuro-behavioural paradigm.

There are several possible reasons to account for why there were no significant changes observed from pre- to post-treatment on measures of cognitive flexibility (DCST) and fine motor dexterity (pegtest). The NIH toolbox tests were novel in their application to children with TS. The DCST and pegtest may not have been sensitive to change in this population. This study was also underpowered to detect small effects and it is also possible that treatment effects were diluted by group delivery; however it is unlikely that there was any meaningful clinical change given similar performance at pre- and post-assessment in both groups on these measures. Finally, there is much debate with regards to whether executive functioning is in fact impaired in children and adults with TS (e.g. Lin, Lai & Gau, 2012). In the present study there was a wide range of individual baseline performance on the DCST. At baseline, performance on the DCST across both groups ranged from the 5th to 95th percentiles. This measure therefore did seem successful in capturing a full spectrum of neuropsychological profiles and it was not the case that all children were simply unimpaired and unlikely to demonstrate change.

Further investigation of the relationship between performance on measures of neuropsychological functioning and tic severity

The findings in this study were mixed. The DCST and Flanker tests were tentatively indicated to be most sensitive to change in this population and most closely linked with tic severity, demonstrating large effect sizes. However, some positive associations between changes in tic severity and fine motor skill on the pegtest were also found. Many associations did not survive Bonferroni-correction. This may be linked to the present study being underpowered, or the result of multiple comparisons inflating the Type I error rate.

The finding that baseline scores on the Flanker test were significantly and negatively associated with tics observed at baseline (r = -0.48) was expected, given that the Flanker was the only test to demonstrate change in the primary analysis. It also supports the view that changes on the Flanker test in the primary analysis may not have been due to practice effects. However, the findings must be interpreted with caution given that no significant associations were found on the YGTSS and the tic observation measure had questionable inter-rater reliability and unclear blinding success. Pre-post changes on the YGTSS were found to positively correlate (r = 0.67) with changes on the DCST, which in many ways tests similar aspects of neuropsychological functioning such as attention and response inhibition.

Predictive relationships

As a result of analysing the CBIT group only, there was a sizeable loss of power for predictive analyses, limiting the ability to detect small or medium effects. However, it was still considered of value to conduct these analyses to inform future studies.

For children in the CBIT group a significant positive association was found between baseline fine motor skill and the amount of change on YGTSS motor subscale scores from pre- to post-treatment. This provides some support for the prediction that children with better neuropsychological functioning at baseline may benefit most from CBIT, demonstrating a large effect size (r = 0.82) and surviving Bonferroni correction. This finding echoes that of Bloch and colleagues (2006), who demonstrated strong links between a more fluent performance on the Purdue Pegboard test and reduced tic severity. It is also interesting that both measures were related very specifically to motor function. Causation cannot be implied from associative relationships; however it is possible that better fine motor dexterity skills at baseline were reflective of less dysfunctional neural circuitry underlying motor tics (Wang et al., 2011). A tentative suggestion may be that children with strong motor skills may have been better able to engage with the CBIT program and master the techniques.

In the present study ADHD symptomatology did not appear to be associated with, or predict, CBIT treatment outcomes, as previously found by McGuire and colleagues (2014) using a moderator analysis. Non-significant findings may have been the result of the range of ADHD symptomatology being too narrow. Only 18% of children in the CBIT group surpassed the clinical threshold for ADHD-hyperactive/impulsive subtype. There was however a higher range of ADHD-inattentive symptomatology, with 59% of children in the CBIT group surpassing the clinical threshold. The range of ADHD-inattentive symptomatology observed was more representative of international ADHD in combination with TS prevalence levels, which have been estimated at 60% (Freeman et al., 2000).

Strengths and limitations

This study had many strengths and weaknesses, which are summarised in Table 10. Key points will be briefly discussed; however are examined in more depth in the Critical Appraisal. The main strengths of this study were that it followed a randomised and controlled, single-blinded design, using highly protocol-driven assessment procedures and fully manualised interventions. The primary tic severity outcome measure (YGTSS) had excellent psychometric properties and is considered the gold-standard measure in both research and clinical settings. An intention-to-treat analysis was used to examine pre- and post-intervention data.

One main limitation of this study was the absence of waiting list or individually-delivered CBIT control groups. These limitations made it difficult to understand active treatment components relating to the group format, as well as to compare outcomes with naturally-occurring changes over time, thus limiting possible conclusions. Other limitations included the small sample size (which rendered the study underpowered to detect small to medium effects), questionable inter-rater reliability on tic severity measures and unclear validity of direct tic observation measures. Lastly, children received treatment as usual in terms of medication management and school liaison,

which can also be considered treatments for tics and may blur the effects of the treatments offered. Levels of school liaison work conducted by the clinical team for each participant were not monitored during the study.

Table 10: Strengths and weaknesses of the present study

	Strengths	Weaknesses
Design	Randomised and controlled experimental design; Blinding of researchers and raters; A repeated-measures approach allowing participants to act as their own controls; An active control group that was similar in structure, frequency, duration and therapist contact; Neither group was portrayed as superior	No waiting-list or individually-delivered CBIT control groups; Blinding of researchers was not assessed for raters of the tic observation videos; Children and parents could not be blinded to group intervention
Sample	A mixture of boys and girls attended the groups, presenting with a wide range of tic severity, tic-related impairment, co-morbid difficulties and medication statuses; The sample was indicated to be representative of clinical populations that declined participation in the study, according to age and gender variables; Children were recruited from a diverse range of geographical locations across England	Small sample (n = 33); No participants had chronic vocal tic disorder; Clinical opportunistic sample
Setting	Specialist national TS clinic; Staff designing and running the groups were highly experienced in working with TS and delivering CBIT; Real-life clinical setting increases generalisability to clinical practice	Children seen in the national specialist TS clinic may not be representative of those seen in primary care, other paediatric or clinical settings, or in the community; Single-centre setting
Measures	Excellent psychometric properties of the primary tic severity outcome measure (YGTSS); Combination of direct and indirect measures of tic severity; The NIH toolbox tests were administered in a standardised format via a computerised program, which measured reaction times to the accuracy of 100th of a second and reduced the likelihood of human error.	Direct tic observations were not well-validated measures - They were also unable to capture tics in the lower body, or quiet phonic tics; The scoring protocol for tic observations was complicated and inter-rater reliability was questionable; One month testing periods for pre- and post-assessments could be considered too lengthy, potentially impacting on performance and outcomes; Tics naturally fluctuate, thus using only two singular time points is not ideal as it may not be a reliable representation of tic severity; Medication changes were only measured by asking parents retrospectively at the end of treatment.
Interventions	Both group interventions and all parent sessions were structured and fully manualised; Fidelity to manual content was 100% in all child groups; The study was highly protocol-driven with regards to assessment and scoring procedures; All child groups were led by the same senior colleague with extensive experience in treating TS.	The September groups had a one week mid-treatment break whereas the November groups had a three-week break; Children received treatment as usual with school liaison and medication, which can also be considered treatments for tics; Fidelity to the manual in the parent groups was not assessed.
Statistical analysis	Intention-To-Treat (ITT) analysis tested the effectiveness of group interventions as originally allocated, including non-attenders and those lost to follow-up; Separate analyses for attenders-only and excluding potentially confounding variables or cases.	ITT analysis is more susceptible to Type II error and is very conservative; This study was underpowered due to a small sample size, particularly for analyses using the CBIT group only (n = 17), making it more difficult to detect small to medium effects. Practice effect data for the NIH toolbox tests was unavailable.

Conclusions and clinical implications

There is tentative evidence to support both CBIT and PE group interventions as feasible and effective, in terms of improving tic severity and inhibitory processes of

neuropsychological functioning. CBIT was found to be superior in reducing motor tic severity. Children with various levels of ADHD symptomatology were able to engage with CBIT. The relatively low rates of tic worsening following the interventions, as well as positive feedback from parents, should reassure other parents and clinicians who may be concerned about exposing their child to others with tics. The development of treatment manuals for these group interventions will support their eventual dissemination to the wider clinical and research communities. The development of local provisions will be vital given that a large number of families declined participation due to distance to travel. The findings from this study have the potential to expand treatment options for children with TS and their families.

Future research

A one year follow-up assessment of these groups is planned, to assess the durability of treatment gains in the long-term. Additional recommendations for future research are considered within the context of plans to conduct a larger-scale RCT, which will ideally use multiple sites. A larger sample size would increase statistical power to detect small and medium effects. This would also permit a further examination of predictive factors. A five-armed design would be advisable if enough children are recruited, in which wait-list, individual therapy and parent-only control groups would be added to further examine active treatment components. If resources are limited, an extra assessment time point of three months before the groups could be added to the current design, in which children would act as their own wait-list control (two-month period, equivalent to group duration).

It would be of interest to evaluate the impact of extending the duration of the groups. This would allow an examination of whether additional sessions could combat any possible diluting effects of group-delivery on treatment outcomes, raising tic reductions towards levels observed from individually-delivered CBIT. All three measures of neuropsychological functioning appear worthy of further investigation

and should be included in future assessment protocols. A multisite approach for the larger-scale RCT would be beneficial to demonstrate treatment effects outside of the specialist TS clinic, which if successful could allow these group treatments to be more widely accessible.

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Part 3: Critical Appraisal

Introduction

The American Psychological Association (APA) Presidential Task Force on Evidence-Based Practice (2006) has reported "psychology's fundamental commitment to sophisticated evidence-based practice" (pp. 271). Randomised Controlled Trials (RCTs) are widely considered the gold-standard experimental design. They have been cited as the most appropriate research design for evaluating the efficacy or effectiveness of standardised clinical interventions compared with a placebo or alternative intervention (APA Task Force, 2006) and establishing a psychological intervention as empirically supported (Chambless & Ollendick, 2001). For clinicians and researchers embarking on the quest to carry out an RCT, some challenges such as cost and feasibility have already been highlighted in the literature (e.g. Stephenson & Imrie, 1998). This critical appraisal allows the opportunity for further reflections on the challenges encountered throughout the process of conducting a moderately-sized pilot RCT, from the early design and recruitment stages through to data collection and analysis. Combined with this in each section will be a further discussion of the strengths and weaknesses of the study.

Design

Strengths and weaknesses

This study followed a randomised and controlled experimental design. While conclusions were limited due to the absence of waiting list or individually-delivered CBIT control groups, the active control (psycho-educational) group allowed treatment outcomes to be compared when controlling for group context and therapist contact factors within a structured and interactive environment. The repeated-measures design allowed children to act as their own controls from pre- to post-treatment. It also reduced the required sample size to achieve adequate statistical power.

Researchers conducting all pre- and post-assessments were successfully blinded to treatment allocation, minimising bias during data collection and scoring. Blinding success was not however evaluated for raters scoring the direct tic observation data. This is unfortunate and it was realised in hindsight following the completion of the study, although it could be considered of relatively less importance given that the Yale Global Tic Severity Scale (YGTSS: Leckman et al., 1989) was considered the primary tic severity outcome measure. It was not possible to conceal treatment allocation from parents and children, who subsequently may have been subject to expectancy effects. This was balanced in part by both treatments being portrayed as having potential benefits, with neither treatment being superior. This was endorsed on written information sheets as well as by the researchers and staff. The expectation that children may improve by attending either group limited the potential for experimenter effects. On a subjective level, many families did appear to favour the CBIT group and the psycho-education groups had higher levels of non-attendance. Thus, expectancy and motivational biases may have occurred.

Discussion

From conception this project was envisaged as a joint piece of work, acknowledging that it would require a huge amount of time and effort, with trainees being involved in all stages from design and recruitment to completion. I undertook this with enthusiasm, driven by a passion to discover more about Tourette Syndrome (TS) and a sense of curiosity, having never been involved in conducting an RCT before. The original design for the project was very ambitious and involved a 3-armed design for an RCT, in which a waiting list control group was included, aiming to achieve a sample of n = 60. Waiting list controls were originally planned to be a separate group, however an alternative suggestion was that children could be tested at three time points (e.g. three months before, one month before and one month after the groups) and act as their own controls throughout. The proposal for this RCT was however

described by a UCL reviewer as a “herculean” doctoral project, thus it was decided to conduct a pilot RCT aiming for a slightly smaller sample. The decision had to be made jointly, between both trainees and supervisors, regarding whether to cut the waiting list or psycho-educational control groups, due to limited resources (primarily trainee time commitments) and the need to scale down the study. It was felt that there was a stronger ethical argument towards providing children with an active alternative treatment. Thus, the final design did not include a waiting list control group. In these early stages I found it particularly helpful to collaborate with the other trainee and utilise the expertise of their supervisor, as well as my own supervisor, through several joint planning meetings.

One aspect of the project that was considered vital was that it had high external validity and was reflective of real-life clinical samples and settings, in order to be generalizable to clinical practice. For this reason, it was agreed that children would continue to receive treatment as usual in terms of school liaison and medication. While it was acknowledged that this would inevitably compromise internal validity and could potentially introduce confounding factors in the treatment of tics, it was also important as many children were on medication and excluding them would have significantly limited recruitment. Medication factors were also controlled for as much as possible in statistical analyses.

Interventions

Strengths and weaknesses

All parent and child groups were fully manualised. This standardisation was essential to ensure that the different rounds of groups in September and November were consistently delivered, thus allowing the data for CBIT and psycho-educational groups to be merged. In the child groups, which were all led by the same highly experienced Consultant Clinical Psychologist, fidelity to the manual was 100% which reaffirmed

this consistency. The group-based CBIT child groups were comparable to individually-delivered CBIT protocols led by Piacentini and colleagues (2010) in terms of the length of group sessions (2 x 90 minutes; 6 x 60 minutes) and the number of sessions (8). However, Piacentini and colleagues (2010) conducted sessions over 10 weeks, with the last two sessions occurring biweekly. In the present study, it was necessary to provide the September groups with a one-week mid-treatment break during half-term and the November groups with a three-week Christmas break. There are no known studies examining the effects of mid-treatment breaks of different lengths and this did introduce variation that was not controlled for. This also meant that the groups differed slightly in terms of structure compared with individual treatments.

Fidelity to the manual was not monitored for the parent groups, which although not the main focus of the study were an important aspect of the treatment package.

Discussion

While there is much debate among clinicians regarding the use of manualised treatments (Addis, Wade & Hatgis, 1999), the clinical team for this project were enthusiastic and dedicated to manualising these group interventions. The manuals were carefully designed, primarily by the Consultant Clinical Psychologist with input from the clinical team and both trainees, so that they were clearly written and thorough yet easy to follow. Staff were familiar with the importance of following the structure of the manual, whist simultaneously building strong therapeutic relationships and individualising examples and discussions to maximise engagement.

Upon taking on the project, I had assumed that all treatments had already been manualised. This process in fact took several months and required collaboration between a number of staff members to complete. I was required to suggest a plan for one session (ADHD) to bring the psycho-educational groups up to eight sessions,

from the original six that had been run at the hospital for many years (Murphy & Heyman, 2007). I found this a challenge and spent some time completing the task, having rarely worked with children with ADHD before; however I learned a lot in the process and was pleased to contribute to the groups. The manualised group treatments were ultimately written to a very high standard, although the time and energy commitments associated with this understandably added an element of pressure within the context of a time-limited, high intensity doctoral project.

It is unfortunate that an assistant psychologist was not available to measure fidelity to the manual in the parent groups. It was not considered appropriate to ask clinicians to tick off the manual checklist themselves, as there were concerns that this might interrupt the flow of the sessions and impede their ability to concentrate on running the groups.

Measures

Strengths and weaknesses

This study combined both direct and indirect measures of tics to ensure a robust assessment of tic severity. The YGTSS is a well-validated measure which has excellent psychometric properties. It allows a retrospective evaluation of tic severity on a number of scales regarding the previous week, incorporating input from children and parents. This is essential as at times parents may notice tics that children are not aware they are doing, and vice versa, children may experience more subtle tics that parents do not notice. Children may suppress tics when being observed, or tics may worsen due to anxiety or exhaustion. Tics are also known to fluctuate ("waxing and waning"). When measuring tics at only two single time points it was essential to use a measure such as the YGTSS which allowed for the consideration of tic severity over a slightly longer time period, to ensure valid and representative measurements were obtained. The direct observational measurements of tics used in this study were

highly protocol-driven and standardised; however scoring protocols were complex, lower body or quiet phonic tics could not be captured and inter-rater reliability was questionable. The YGTSS also had questionable levels of inter-rater reliability, which limits the confidence from which conclusions can be made using these measures.

The National Institutes of Health (NIH) computerised measures of neuropsychological functioning (www.nihtoolbox.org) were well-validated measures, which minimised the likelihood of human error and allowed highly accurate recordings of reaction times.

Discussion

The YGTSS and Premonitory Urge for Tics Scale (PUTS: Woods et al., 2005) were measures which had been commonly used within the TS Clinic, which was the setting for this study, thus being natural choices. However, the process of choosing and implementing additional measures was not quite so straightforward.

Very few pre-existing protocols were identified which could provide standardised assessments of video-recorded direct tic observations. Two were examined in detail. The first was a protocol by Goetz and colleagues (1987), which involved filming people with tics in different positions (full frontal body, far; head and shoulders only, near) during different conditions (relaxed with and without the examiner in the room), using two cameras. This procedure was considered fairly complex and the researchers in the present study did not have access to two cameras, thus moving to the second protocol (Himle et al., 2006). Although we were still not able to follow this protocol fully, it only required one video camera and was considered the best fit for this study. Children were able to watch a television show whilst being filmed, which was considered practical for researchers during an already full testing schedule, as well as being enjoyable for the child. We had limited resources and instead of using a video-camera were able to download camera software to laptops. This created some complications which initially needed to be solved before testing, such as adjusting

settings so that all confidential video data would save directly to an encrypted hard-drive within each laptop.

This measure initially seemed a relatively simple task, involving the filming and counting of tics observed. However, following the protocol of only counting previously-defined tics (from the tic list for each child, as in Himle et al., 2006) was surprisingly difficult, with children frequently presenting with severe and enduring bouts of simple and complex tics. Some of the recordings were not ideal, with sounds being inaudible or children moving out of the video range, eating or fidgeting. It had originally been intended that both trainees conducting all pre- and post-assessments would score this observational data. However, due to the immense demands of the project it became necessary to accept support from post-doctoral colleagues on the clinical team with regards to scoring. While this was intended to ease pressure and time commitments for the trainees, it also resulted in the need to develop a detailed written scoring protocol and to provide them with training in how to score the videos, which ultimately became very time consuming in itself.

In terms of measures examining neuropsychological functioning, initially paper-based measures, or measures that had already shown indications of being sensitive to differences or change in people with TS, were strongly considered (e.g. Stroop test: Marsh et al., 2007; Purdue pegboard test: Bloch et al., 2006). However, these tests were often expensive or bulky to transport. The NIH online tests were freely available to use, well-validated, nationally normed and considered appropriate for the study research questions by the trainees and supervisors. However, what was intended to be a relatively simple measure to use ended up being very difficult to set up. Guidelines to facilitate set up were complex and as the service was free, support services (based in the US) were difficult to access. Ultimately, extra equipment had to be purchased and it took the researchers several months to learn how to set up and administer the tests. We also encountered issues with internet access in the hospital

and also in some homes, due to inadequate wireless internet speeds or bad weather. We purchased dongles for these circumstances, which provide portable internet access; however these did not provide adequate speeds for the NIH test to accurately record reaction times and were thus not used.

Setting

Strengths and weaknesses

The groups were conducted within the context of a highly specialist TS clinic in a national hospital for children. This service is likely to attract children with more severe and complex difficulties, who may not be representative of children seen in other primary care, paediatric or community settings. The groups were run as part of routine clinical practice, increasing their relevance and generalisability to real-life clinical settings. Staff designing and delivering the groups were skilled clinicians within the area of TS and CBIT interventions. Less experienced staff in different settings may not observe the same treatment outcomes as those in the present study, thus limiting the generalisability of the results until a multi-centre trial is conducted. Assessments were primarily conducted within families' homes, which although convenient for the families was less of a controlled environment and introduced some elements of variability. This was controlled for as much as possible by requesting a quiet room with a table and chairs in every assessment.

Discussion

It was a privilege to be involved with these groups which were led by such experienced and knowledgeable staff within a high-profile setting. The other trainee was on clinical placement within the department for the first six months of the study, while I was on clinical placement in the clinic for the final year of the study. It was extremely helpful in some ways that one of us was always based in or near the clinic, so that the large number of planning meetings, study preparation and recruitment

sessions could be organised on site. However, it was often important to clarify clinical and research roles, separating the two when possible due to staff or patients becoming unclear (e.g. reminding parents in the study that although we worked clinically within the department our involvement with them was in a research capacity). Research work (preparation, recruitment, scoring, data entry) was often done at the end of a long clinical day. This amounted to many hours in the same setting, which although very convenient made it difficult to create a mental and physical separation to allow breaks when switching between research and clinical roles. Working within the clinic and in close proximity to where the groups were held, I and the other trainee had to work very hard to remain blinded to treatment allocation, with the support of the clinical team and families.

The decision was made that we should offer to conduct all assessments at families' homes for their convenience, particularly in light of being unable to fund their travel to and from the groups. Fortunately we were able to secure funding for this from a charity named Tourette's Action. The home environment wasn't ideal, with siblings, pets, small tables, distractions and slow or interrupted wireless internet signals providing challenges to testing. However, for the majority of assessments we were able to establish a quiet environment in which children were not disturbed during testing.

Preparation

Discussion

The stages of preparation for this project were numerous and time intensive. Firstly, all detailed child and parent information sheets, consent forms and GP letter drafts needed to be written. We had few examples to base these on and their design and content took a considerable amount of input to complete. One service-user (a mother of a boy with TS) was involved in the design and preparation stage of the study.

Ideally we would have liked to involve more adult and child service-users, however time was of the essence and unfortunately this was not possible.

Ethical approval first had to be sought from UCL and Royal Holloway, University of London, then by the hospital's Clinical Research Adoption Committee and finally by the London Queen Square Research Ethics Committee. All stages required lengthy proposals, the latter two of which involved full panel questioning to gain approval for the wider project as a whole, requiring careful collaboration with the other trainee and the clinical team. This process took over six months, leading to more pressure to recruit families quickly before the end of the summer and the start of the first set of groups.

Regular planning meetings were held with the clinical team, both trainees and our supervisors. There were then numerous other tasks, which included: The manualisation of all treatments, application for additional graduate funds, ordering of equipment, development of highly detailed written testing protocols for pre- and post-assessment visits, practicing and mastering the application of all testing batteries and protocols, developing scoring protocols for the direct tic observation measures and instructions for randomisation, training post-doctoral colleagues in scoring, creating visual timetables, developing the demographic questionnaire to be used at intake and creating large databases for data entry. It was also necessary to register the trial with an International Standard Randomised Controlled Trial Number (ISRCTN; 50798741) via the National Institute for Health Research (NIHR) Portfolio Database (http://www.controlled-trials.com). This again took some time and the NIHR Comprehensive Clinical Research Network required regular updates regarding progress at each stage of the study.

Recruitment, sample and randomisation

Strengths and weaknesses

Thirty-three children in total were recruited, which was considered an adequate sample size for a pilot study. Although the sample included more boys than girls, just under one third of the sample were girls, which could be considered enough to support the generalisability of these findings to both genders. The study took an inclusive approach, using wide-ranging eligibility criteria and few reasons for exclusion. Children with numerous co-morbid difficulties, as well as varying levels of tic severity and tic-related impairment, were accepted. This study was one of effectiveness and not efficacy. It compromised internal validity and the use of "pure" experimental samples by maximising the relevance of the research sample to the target population, who typically present with a range of co-morbidities.

The findings of this study are less applicable to children with Chronic Vocal Tic Disorders, as no children in the recruited sample were given this diagnosis. Participants were from a range of socioeconomic and geographical backgrounds, which is unusual for a single-centre setting and reflective of the national service offered by the clinic. This study followed a clear and replicable randomisation protocol, conducted by a member of staff who was involved in the running of the groups but not in the research project or the scoring of measures.

Discussion

Initially, 153 families were contacted and assessed for eligibility by the other trainee and I. This typically involved many hours of evening calls from the hospital, as most families were not available during working hours. It was necessary for us to purchase separate mobile phone SIMs for the study in order to be available for families to contact at any time to discuss the study. We were surprised that some families who lived up to three hours from London were very willing to take part in the study. This presented us with a dilemma as it required a significant amount of travel to conduct assessments, however we not over-subscribed and were reluctant to deny children the opportunity of participating for this reason.

Some families objected to randomisation and expressed a clear preference as to which treatment their child should receive. These families were reminded of the requirements of randomisation and that they would need to be willing to accept either treatment if they were enrolled into the study. Some families subsequently declined participation and some accepted; however preferences were not monitored and could have been a source of bias in terms of motivational and expectancy effects. Great consideration was given regarding when the groups should start, using previous experiences of the team who reported that groups over the summer holidays tend to have low attendance rates.

This study followed a protocol in which children were randomised sequentially, allowing groups to commence as soon as enough children were recruited. This was essential given time constraints. Randomisation could have been stratified by medication, however it was already stratified by age and gender so it was considered preferable to control for medication changes or types (i.e. stimulant versus non-stimulant) in the analyses.

We were responsible for printing, packing and posting all paper-based documents, such as information and consent sheets as well as questionnaires, if families agreed to take part. This was time-consuming and required strong organisational skills.

Assessment procedures

Strengths and weaknesses

All pre- and post-assessments were highly protocol-driven, involving a comprehensive battery of computerised tests, written questionnaires and interviews with parents and children. NIH computerised measures of neuropsychological functioning provided researchers with standardised instructions to follow, along with a structured testing procedure. This ensured consistent delivery between the two trainees conducting all assessments. Trainees spent many hours discussing and

practicing the administration of all measures prior to testing, to ensure agreement on scoring procedures.

One weakness of the assessment procedures was that the one month testing periods for pre- and post-assessments could be considered too lengthy. This study did not include an analysis of the effects of shorter or longer durations of pre- or post-assessment periods, although did control for those that were longer than one month in all statistical analyses. Medication changes were also only measured by asking parents retrospectively at the end of treatment, which was subjective and dependent on memory.

Discussion

In total each trainee conducted roughly 33 assessments each. This proved a taxing schedule, given that most visits took half a day or a whole day and involved travelling around the country with a large suitcase of testing equipment. Throughout this process, high levels of organisation and commitment were essential. I was very grateful for the support of the other trainee. Joint working relieved time and resource pressures, as well as offering a sense of personal support through a long and intensive project. We maintained good communication to ensure the smooth running of all testing phases and were able to follow Lone Working procedures by arranging a contact time to check in with each other following each home visit.

Upon taking on the project, we had originally intended to complete testing sessions on weekdays and only occasionally on weekends. However, ultimately most visits were required to be on weekends. This was because testing needing to be conducted in the home environment with minimal impact on schooling and many families were away during the summer holidays or half-term. It was not felt appropriate or ethical to test children in the evenings after school due to high levels of fatigue and the need for rest. We recognised that testing periods of one month before and after the groups

could be considered quite lengthy. However, this was unavoidable given the time-intensive testing protocols and there being only two of us to conduct the assessments. Each trainee conducted pre- and post-assessments with the same children, which was beneficial in terms of reliability of scoring as well as enabling the building of rapport and returning to familiar surroundings on the second visit.

Data analysis and writing up

Strengths and weaknesses

This study was underpowered due to the small to moderate sample size obtained, particularly for associative analyses with the CBIT group only, making it more difficult to detect small to medium effects. It applied an Intention-To-Treat (ITT) analysis as the primary data analytic approach. While this approach can be criticised for being highly conservative and vulnerable to Type II error, it has significant benefits in testing the effectiveness of group interventions in relation to original treatment allocations. ITT analysis also minimised the impact of non-attenders and those lost to follow-up on statistical power. Statistical analyses procedures were thorough, including separate analyses excluding potentially confounding data and for attenders only, in which outcomes were evaluated when the full treatment protocol was adhered to. The absence of practice effect data for the NIH tests and waiting list controls led to an inability to fully support the idea that observed improvements were based on the group intervention, as opposed to being the product of time.

The study was conducted and written up in line with the CONSORT guidelines for non-pharmacological randomised controlled trials (Boutron et al., 2008), which consists of an evidence-based checklist and flow diagram of recommendations for reporting RCTs with the highest level of accuracy.

Discussion

The analytic approaches used within this study took some consideration, given the large number of hypotheses and the variety of ways in which the data could be analysed and presented. It was also quite a challenge to write up this large project in line with the comprehensive CONSORT guidelines, whilst keeping in mind the need for the project to be written succinctly and in keeping with the limitations of a D.Clin.Psy project. The CONSORT guidelines required the inclusion of many additional sections such as randomisation, blinding and adverse effects, which added to this difficulty. Given the typically high impact of RCTs, one feels a sense of responsibility to provide a report of the highest quality and accuracy. However even in the outside world of research and publications, there will always be limits to adhere to and this can be a challenge for researchers, to ensure that findings are presented in comprehensive yet accessible forms.

Conclusions

In summary, while this project was demanding and intensive in terms of time and resources, it was highly rewarding and allowed much room for learning and striving towards the highest standards within a centre of clinical excellence. This project would not have been possible without effective joint working, the support of an active and dedicated clinical team and committed research supervisors.

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Appendices

Appendix 1: Structured process for making exclusions during initial screening process (following database searches)

A flowchart process was followed starting from top to bottom, to exclude studies that were not eligible for this review:

- ➔ Not a journal (e.g. reviews, posters, commentaries, letters)
- ➔ Not human (i.e. animal studies)
- ➔ ADHD not primary disorder being investigated [studies not to be included if co-morbid disorders are in the title and appear of equal interest to ADHD e.g. "ADHD and Conduct Disorder" – General sample with comorbidities is acceptable]
- ➔ ADHD not diagnosed (e.g. at risk only or symptoms at a non-clinical level)
- ➔ Not a treatment intervention
- ➔ Not a psychological intervention (e.g. medication, homeopathy, neurofeedback, physical training, sensory stimulation, acupuncture, dietary, etc)
- ➔ Not children aged 5 – 18 years (i.e. pre-schoolers or adults)
- ➔ Parenting intervention only
- ➔ Teacher intervention only
- ➔ Parent and teacher interventions only
- ➔ Family interventions only
- ➔ Clearly not groups (i.e. individually-delivered sessions clearly stated; single case studies; telephone delivery)
- ➔ Summer treatment programs
- ➔ Social skills training only

Appendix 2: Database search terms with Boolean operators

MEDLINE:

attention deficit.mp
exp Attention Deficit Disorder with Hyperactivity/
attention-deficit.mp
hyperactiv*.mp
hyperkin*.mp
adhd.mp
addh.mp
adhs.mp
1 or 2 or 3 or 4 or 5 or 6 or 7 or 8
child*.mp
boy*.mp
girl*.mp
teen*.mp
adolescen*.mp
youth*.mp
school-child*.mp
schoolchild*.mp
young*.mp
10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18
group*.mp
exp Group Processes/
exp Psychotherapy/
20 or 21 or 22
interven*.mp
exp Treatment Outcome/
treat*.mp
technique*.mp
train*.mp
program*.mp
impact*.mp
therap*.mp
outcome*.mp
trial*.mp
24 or 25 or 26 or 27 or 28 or 29 or 30 or 31 or 32 or 33
18 and 19 and 23 and 34
limit 35 to journal article
limit 36 to english language
limit 37 to yr="2007 -Current"
limit 38 to humans
limit 39 to "all child (0 to 18 years)"

PsycINFO:

attention deficit.mp.
exp Attention Deficit Disorder with Hyperactivity/
exp Attention Deficit Disorder/
exp Hyperkinesis/
attention-deficit.mp.
hyperactiv*.mp.
hyperkin*.mp.
adhd.mp.

addh.mp.
adhs.mp.
1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10
child*.mp.
boy*.mp.
girl*.mp.
teen*.mp.
adolescen*.mp.
youth*.mp.
school-child*.mp.
schoolchild*.mp.
young*.mp.
12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20
group*.mp.
exp Group Counseling/
exp Group Intervention/
exp Group Psychotherapy/
22 or 23 or 24 or 25
interven*.mp.
exp Treatment/
exp Behavior Therapy/
treat*.mp.
technique*.mp.
train*.mp.
exp Training/
program*.mp.
impact*.mp.
therap*.mp.
exp Psychotherapy/
exp Cognitive Behavior Therapy/
outcome*.mp.
exp Psychotherapeutic Outcomes/
trial*.mp.
27 or 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41
11 and 21 and 26 and 42
limit 43 to peer reviewed journal
limit 44 to english language
limit 45 to yr="2007 -Current"
limit 46 to human
limit 47 to (100 childhood <birth to age 12 yrs> or 200 adolescence <age 13 to 17 yrs> or 320 young adulthood <age 18 to 29 yrs>)

EMBASE:
attention deficit.mp
exp attention deficit disorder/
attention-deficit.mp
hyperactiv*.mp
exp hyperactivity/
hyperkin*.mp
exp hyperkinesia/
adhd.mp
addh.mp
exp minimal brain dysfunction/
adhs.mp

1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11
child*.mp
exp child psychology/
boy*.mp
girl*.mp
teen*.mp
adolescen*.mp
youth*.mp
school-child*.mp
schoolchild*.mp
young*.mp
13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22
group*.mp
exp group psychology/
exp group therapy/
24 or 25 or 26
interven*.mp
treat*.mp
technique*.mp
train*.mp
exp training/
program*.mp
exp program effectiveness/
exp program efficacy/
impact*.mp
therap*.mp
exp therapy/
outcome*.mp
exp treatment outcome/
trial*.mp
28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41
12 and 23 and 27 and 42
limit 43 to journal
limit 44 to english language
limit 45 to yr="2007 -Current"
limit 46 to human
limit 47 to (preschool child <1 to 6 years> or school child <7 to 12 years> or adolescent <13 to 17 years> or adult <18 to 64 years>)

Appendix 3: Quality assessment components and ratings for the EPHPP (as presented by Thomas et al., 2004)

[**SENSITIVE OR COPYRIGHTED MATERIAL REMOVED; See Thomas et al., 2004 for further information]**

Appendix 4: Data extracted from 9 excluded studies not included in evaluation of effectiveness

Primary author (date)	Time points	Intervention summary	Medication status	Outcome measures	Main findings	Reported effect sizes	Main limitations	Support for groups?
Hinshaw (1984) - Study 1 only	Pre, post	1. Small group training in **cognitive self-regulation skills +** methylphenidate 2. Small group training in cognitive self-regulation skills + placebo	All on stable methylphenidate dose for 3m prior to study (all evidenced to have positive response); Medication then manipulated	*Rating scales during Behavioural Provocation Tests: 1. Global:* a) Self-control b) Intensity c) Provocation strength. 2. *Specific behaviour:* 4 verbal and 5 motoric behaviour categories were scored as present or absent for 6sec observation intervals	Irrespective of medication, both groups displayed significantly better abilities on some global (self-control) and specific measures (vocalising, verbal retaliation, use of purposeful alternative activities) post-intervention. Main effects for medication were found for one global (Intensity) and one specific measure (walking away). No significant interactions between medication and group were found, indicating no advantage for the combination of medication and group cognitive-behavioural intervention.		Weak methodology; Medication/ placebo could account for improvements in both groups; No adequate control group; Possible habituation to provocation at post-test; Small *n*	Some support (short-term), but many limitations
Looyeh (2012)	Pre, post, 1m	1. **Group narrative therapy** 2. Waitlist control	No subjects on medication during or up to 30 days after completion of the groups	*CSI:* Teacher-rated inventory based on DSM-IV criteria, measuring 14 major children's psychiatric disorders including ADHD and related subtypes.	Intervention group showed a significant decline in ADHD symptom scores from pre- to post-test. This was maintained at follow-up. Wait-list controls showed no significant change in ADHD symptoms at post-test or follow-up.	Therapy vs controls, large effect sizes at post-test (d=0.93) and medium at follow-up (d = 0.74)	Weak methodology; Small *n*; One outcome measure; All females	Some limited support (short and long-term)
Miranda (2000)	Pre, post, 2m	1. **Cognitive-behavioral self-control training** 2. **Cognitive-behavioral self-control training + anger management training**	No participants were prescribed medication for ADHD for the duration of the study	*Parent-rated:*1. The Hyperactivity Qur 2. Scale of Behavioral Problems *Teacher-rated:* 1. Abbreviated Conners Rating Scale 2. CTRS (adapted) 3. Problems Inventory in the School (e.g. antisocial behavior, anxiety, inhibition) 4. Self-Control Rating Scale 5. Social Skills Assessment Teacher Form 6. School grades (maths/ language/humanities).	Both interventions produced considerable improvements on many outcome measures (e.g. ADHD symptoms, behaviour, self-control), as rated by parents and teachers, at post-test and follow-up. This was the case whether there was evidence of aggression or not. No significant changes in school grades.		Weak methodology; Small *n* (particularly when divided into aggressive/non-aggressive); No adequate control groups	Good support from outcomes (short and long-term), but limitations

Ozcan (2013)	Pre, post	1. **"I Can Problem Solve" (ICPS) program**	21% of sample on medication; 79% not on medication	1. DBDRS (DSM-IV-TR based, adapted), parent / teacher-rated 2. CBCL (adapted), Teacher-rated	Pre-intervention scores for behavioral and emotional problems (e.g. ADHD symptoms, anxious/depressed, ODD, aggression) were significantly improved after participants received the ICPS program. Children's adaptive functioning (working, behaving, learning, happy) also improved significantly from pre to post-test.		Weak methodology; No control groups; Small *n*	Good outcomes (short-term), but limitations
Sibley (2014)	Pre, post (4 weeks after groups), 5-8 weeks (school data)	1. **Supporting Teens' Academic Needs Daily-Group (STAND-G):** Adolescent behavioural intervention group + parenting skills group 2. Waitlist control group	61% of participants were currently prescribed psychostimulant medication	1. *Academic (calculated weekly):* a) Average assignment grade b) Average grade on tests/quiz's/exams c) % turned in assignments 2. *Satisfaction qurs* 3. *Improvement Rating Scale* (parent ratings of improvement in areas such as accepting limits, routines, etc)	The majority of parents and teens expressed high satisfaction with STAND-G. Those in the treatment group displayed improvements pre- to post-intervention across a range of domains according to self and parent ratings on the Improvement Rating Scale. However, there were no significant increases in objective grades at post-test or follow-up (with the exception of test/exam scores for one group pre- to post-test who were older - *see effect sizes)*, relative to control students.	Test/exam scores for cohort with older teens: Large effect pre- to post- (*d*= 1.02). Post to f-up *d*= 0.00	Weak methodology; Expectancy effects of parents and teens for main significant finding; Small *n;* Possible impact of medication	Some support on subjective measures (short-term only), but limited
Van der Oord (2012)	Pre, post, 2m	1. **Mindfulness training** - Child group + mindful parenting training 2. Waitlist control (within-group, whilst awaiting treatment)	4 children were treated with methylphenidate before the study. Dose was kept stable during waitlist and active treatment.	*Parent measures:* 1. DBDRS (parent and teacher-rated) 2. PSI 3. The Parenting Scale - assesses ineffective discipline styles 4. Mindfulness Attention and Awareness Scale 5. ADHD Rating Scale (includes parent's own symptoms)	Significant reduction of parent-rated ADHD behavior of themselves and their child from pre-to post-test and at follow-up. Teacher ratings did not indicate any significant effects of the treatment group.	Pre-Post child ADHD outcomes: Medium - large (*d*=.56-.80). Follow-up similar (*d*=.59-.80)	Weak methodology; Expectancy effects (parents); Small *n;* No active control group	Some support (short and long-term), but limited

Van de Weijer-Bergsma (2012)	Pre, post, 2m, 4m	**Mindfulness training** - Child group + mindful parenting training	One participant was on methylphenidate at time of recruitment. Dose was kept stable throughout the study.	*Parent/teacher:* 1. CBCL (parent only) 2. Teacher Report Form 3. BRIEF 4. PSI - short version 5. Parenting Scale (discipline styles) *Child:* 1. Youth Self Report (behaviour problems) 2. Mindful Attention and Awareness Scale (also parents) 3. Flinders Fatigue Scale 4. Subjective Happiness Scale 5. Amsterdam Neuropsychological Tasks (visual and auditory sustained attention)	a) After mindfulness training, adolescents showed significant improvements on the number of false alarms on an objective measure of sustained auditory attention. This reduced to borderline significance at follow-up. b) At 8-week follow-up only, fathers and adolescents (not mothers), reported significant improvements in executive functioning (BRIEF) and attention problems. c) Directly after training and at d) 8 week follow-up, externalizing problems reduced significantly as reported by fathers, but not by adolescents, mothers or teachers. No significant effects on mindful awareness (of adolescents or parents), fatigue, happiness and internalizing behaviour problems were found at post-test or follow-up.	a) *d*=0.5 (medium) b) *BRIEF* Fathers: *d*=0.6-1.8 (medium-large) *Attention* Fathers: *d*=1.5 (large) Youth: *d*=0.9 (large) c) *d*=0.2 d) *d*=0.3 (Small)	Weak methodology; Expectancy effects (parents and children/ teens); Small *n*; No control group; Possible practice effects for sustained attention task	Mixed findings with some support (short and long-term), but limitations
Pfiffner (2011)	Pre, post	**Collaborative Life Skills Program (CLS) - Child skills group training +** Parent training + Teacher consultation/ classroom component: Adapted from the clinic-based CLAS Program (*see Pfiffner, 2011*) for a school context.	Only two participants (5%) were taking medication for ADHD when recruited. This was required to remain stable throughout the study.	*Parent/teacher:* 1. CSI (ADHD criteria included) 2. Social Skills Improvement System (SSIS), Problem Behaviors Scale (internalizing/externalizing problems) 3. SSIS Social Skills Scale 4. Homework Problems Checklist (parents) 5. Academic Competency Evaluation Scale (teachers) 6. COSS 7. Satisfaction qurs - child/parent	Significant pre- to post-treatment improvement at school and at home in 1. ADHD symptoms 2. Problem behaviors (home only) 3. Social skills (home only) 4. Organizational skills 5. Homework problems (parent measure) and 6. Academic competency (teacher measure). Support for the effectiveness of this adapted treatment in improving student outcomes in a school context.	Med-large: 1. Parent: *d*=0.83; Teacher: *d*=1.19 2. *d*=0.68 3. *d*=0.49 4. Parent: *d*=0.78; Teachers: *d*=0.55 5. *d*=0.96 6. *d*=0.56	Weak methodology; No control group; Small *n*; Expectancy effects; No follow-up	Good support (short-term), but limitations

Pfiffner (2013)	Pre, post	**Collaborative Life Skills Program (CLS) - Child skills group training** + Parent training + Teacher consultation/classroom component: *Extended from Pfiffner, 2011*	4 children (7%) were taking medication for attention or behavior concerns when recruited. This was required to remain stable throughout the study.	*Parent/teacher:* 1. CSI (ADHD criteria included) 2. Homework Problems Checklist (parents) 3. Academic Competence Evaluation Scale (teachers) 4. COSS *Child:*1. School grades (language arts / maths) 2. Woodcock Johnson Tests of Achievement (maths and reading fluency/ comprehension) 3. Classroom behavioral observations (3 occasions over 2 weeks, 15mins each)	Significant pre- to post-intervention improvement at school and at home was found for all measures including: 1. ADHD symptoms 2. Homework problems 3. Organizational skills 4. Academic skills and 5. achievement (Woodcock) 6. School grades 7. Student engagement in classroom. Improvements in organizational skills were indicated to mediate the relationship between improvement in ADHD symptoms and academic skills. Clinically, a large proportion of children showed reliable improvement following the intervention on parent (49%) and teacher (53%) reported ADHD severity. 51% of children were in the non-clinical range for ADHD symptoms at post-treatment.	Med-large (one small) 1. Parent: *d*=1.09; Teacher: *d*=.23 (small) 2. *d*=.89 3. Parent: *d*=.93; Teacher: *d*=.71 4. *d*=.41 5. *d*=.51-.81 6. *d*=.77-1.24 7. *d*=.70	Weak methodology; No control group; Expectancy effects; No follow-up	Good support (short-term), but limitations

Appendix 5: Further information on the neurobiology of tics as reported in a neuroimaging study by Wang and colleagues (2011)

Prior to tic generation the premonitory urge is associated with increased activity in the Primary Somatosensory Cortex (PSC), putamen and amygdala/hippocampus complex (most likely linked with the aversive experience of the PU). Excitatory projections from the PSC to the putamen appear to have a causal influence in triggering tics, sequentially influencing the pallidum and thalamus. The thalamus subsequently activates excitatory projections to the motor cortex. In combination with reduced activity in top-down control areas of these circuits (anterior cingulate and caudate), this leads to disinhibited movements (which can include vocal chords as well as body parts).

Appendix 6: Joint Project: Outline of trainee contributions

Both trainees contributed equally to the design of the study, participant recruitment, data collection and data entry. The author was responsible for consulting the service-user with regards to study design. The other trainee created the Excel and SPSS databases that were used for data entry. The external supervisor was responsible for manualising both groups, although the trainees contributed one session plan each for the additional two psycho-educational sessions that needed to be added. The external supervisor and clinical team ran all of the groups, in which the trainees were not involved (to remain blind to treatment condition). The re-scoring of tic counting videos to test inter-rater reliability was split evenly between the two trainees. The two trainees conducted statistical analyses separately and wrote up their respective thesis projects independently.

Appendix 7: Child end of treatment satisfaction questionnaire

Young Person's Questionnaire for Tourette Group

We would like to evaluate the usefulness of this first group for young people with Tourette Syndrome. It is therefore very helpful for us to get some feedback from you and we would appreciate it if you could fill in the questionnaire. Please also feel free to add any additional comments.

1. **Was the amount of information about the group:**

much too little	too little	sufficient	too much	far too much
☐	☐	☐	☐	☐

2. **Would you have liked the frequency of the group to be:**

Every week	every 2 weeks	every 3 weeks	every 4 weeks	less frequent
☐	☐	☐	☐	☐

3. **Would you have liked the group to take place at:**

10am	12pm	2pm	4pm	6pm
☐	☐	☐	☐	☐

4. **Would you have liked the number of sessions to be:**

2	4	6	8	more
☐	☐	☐	☐	☐

5. **Would you have liked the duration of the group to be:**

1 hour	1 ½ hours	2 hours	3 hours	more
☐	☐	☐	☐	☐

6. **Was the group:**

Not at all enjoyable				Enjoyable extremely
1	2	3	4	5

7. Was the group:

Not at all helpful				Extremely helpful
1	2	3	4	5

8. Were the group leaders:

Not at all helpful				Extremely helpful
1	2	3	4	5

9. What did you enjoy most about the group:

a.

b.

c.

10. What did you learn in the group:

a.

b.

c.

11. What other areas would you have liked covered in the group:

a.

b.

c.

12. Any other comments on how the group could be improved?

Appendix 8: Parent end of treatment satisfaction questionnaire

Parents Questionnaire for Tourette Group

We would like to evaluate the usefulness of this first group for young people with Tourette Syndrome. It is therefore very helpful for us to get some feedback from you and we would appreciate it if you could fill in the questionnaire. Please also feel free to add any additional comments.

Having brought your child to the group and possibly discussed the sessions with your child did you get a sense of:

1. **For you and your child, was the amount of information about the group:**

Much too little	Too little	Sufficient	Too much	Far too much
☐	☐	☐	☐	☐

2. **Would you have liked the frequency of the group to be:**

Every week	every 2 weeks	every 3 weeks	every 4 weeks	less frequent
☐	☐	☐	☐	☐

3. **Would you have liked the group to take place at:**

10am	12pm	2pm	4pm	6pm
☐	☐	☐	☐	☐

4. **Would you have liked the number of sessions to be:**

2	4	6	8	more
☐	☐	☐	☐	☐

5. **Would you have liked the duration of the group to be:**

1 hour	1 ½ hours	2 hours	3 hours	more
☐	☐	☐	☐	☐

6. **Was the group:**

Not at all helpful				Extremely helpful
1	2	3	4	5
☐	☐	☐	☐	☐

7. **Any other comments on how the group could be improved?**

Appendix 9: Measures administered for the wider study

Quality of life (QoL) outcomes for the wider study were assessed using four questionnaires: The Paediatric Quality of Life inventory version 4.0 (PedsQL; Varni et al., 2001), Gilles de la Tourette Syndrome Quality of Life scale (GTS-QoL; Cavanna et al., 2013), the Strengths and Difficulties questionnaire (Goodman, 1997) and a Tourette Syndrome Visual Analogue Scale (VAS). The VAS was developed by TM. It required children to mark points on five horizontal lines, each relating to a different question about the child's experience and acceptance of their tics. QoL measures were included in data collection protocols but were not discussed in this report. Four questions screening for the presence of rage attacks (as included in the Rage Attacks Questionnaire used in Budman et al., 2003) were also distributed to parents in order to characterise the groups, but were not included in the current study.

Appendix 10: Ethical approval letter from UCL

UCL Doctorate in Clinical Psychology

Research proposal review form

Trainee: Katie Edwards
Project title: Group Work for Tourettes syndrome: A pilot study to evaluate the efficacy of a tic-specific behavioural intervention versus psycho-education in improving tic severity and relevant aspects of neuropsychological functioning
Internal supervisor: JK
External supervisor: TM

Reviewer: WM
Date: 20th December 2012

Please indicate one of the following:
__ **Research can go ahead, although some details may still need to be finalised**
__ Proposal needs revision (to be seen again by the reviewer)
__ Proposal seems unsuitable on scientific or practical grounds: will need rethinking

Reviewer's comments:
This proposal describes an ambitious study designed to compare psychoeducation groups and comprehensive behavioural intervention for tics (CBIT) groups with each other. The main outcome measures are tic severity, neuropsychological functioning and premonitory urge awareness. It also aims to test whether certain neuropsychological measures predict response to CBIT treatment. This is a joint project, to be carried out alongside a trainee from Royal Holloway.

This is a revision of a previous proposal, which was excellently written, but which I found to describe a study that was not feasible within the scope of a DClinPsy project. I found Katie's revision to be thoughtful and thorough, and to have engaged with the issues of feasibility that I raised.

I continue to believe this is an ambitious study, that is more risky that most DClinPsy projects. Since the study is reliant on a RCT being carried out at [hospital] within a relatively narrow window of time, the trainees will be vulnerable to delays and hold ups that are outside of their control. However I note that in the revised version of the study, Katie has demonstrated a careful attention to the risks involved and how these might be managed. Therefore, I leave it to her to decide whether she is happy with proceeding with the study in its current form.

I flag up the following for consideration:

1. There was no explicit statement as to how many children there are per group. I assume there are enough to provide 46 participants in four groups, but this should be made explicit. In fact there was some confusion about this, as the timeline suggests the plan is to recruit 40 children; but the power calculation says 46 are needed.
2. Also, the proposal says there are 200 to 250 children who can be approached for recruitment. Is this the number of children seen at [hospital] in the last five years, or is it the number who actually meet the study's inclusion criteria.

3. Please state in the proposal when families will be told which condition of the trial they have been assigned to. I assume this will be after the baseline assessment to avoid any bias due to expectancy effects.
4. I still have some concerns about who will manage the randomisation. Is this going to be part of the assistant psychologist's job? If so it will be essential to ensure that they understand how to randomise effectively, and that there is clarity about their role in the project. Since the trainees are blind to group allocation, they cannot call families to tell them which group they are in, talk families through the process of coming to the group, deal with practical questions about coming to [hospital], deal with the effect of drop out and so on. There needs to be someone at [hospital] to fulfil this role.
5. I did not understand the potential alternative project (Plan B). It seems to involve looking at weekly ratings of tics, homework compliance, attendance and medication status, and analysing how these variables 'interact with co-morbid ADHD and OCD'. Given that there might be recruitment issues with this project, the plan B should be thought through more carefully.
6. I think that an alternative Plan B would be to have an uncontrolled study, just looking at CBIT groups. Participants could have three assessments: (1) Several weeks before commencement of therapy; (2) Immediately before commencement of therapy; (3) Immediately after termination of therapy. This would offer evidence on whether the treatment helps, and what sort of effect size it achieves. Such work could prepare the ground for an RCT.
7. I strongly urge Katie and her collaborators at [hospital] and Royal Holloway to get started on their research as early as possible, to minimise risk of under-recruitment.

I found this proposal to be intelligently written, and to show an impressive rigour. I do not need to see the proposal again, and would like to wish Katie good luck with her study. It has the potential to offer major insights into the treatment of tics.

Appendix 11: Ethical approval letter from Royal Holloway, University of London

[SENSITIVE MATERIAL REMOVED]

Appendix 12: Ethical approval letter from the hospital Clinical Research Adoption Committee

[SENSITIVE MATERIAL REMOVED]

[SENSITIVE MATERIAL REMOVED]

Appendix 13: Ethical approval letter 1 from the London Queen Square Research Ethics Committee

[SENSITIVE MATERIAL REMOVED]

[SENSITIVE MATERIAL REMOVED]

[SENSITIVE MATERIAL REMOVED]

[SENSITIVE MATERIAL REMOVED]

[SENSITIVE MATERIAL REMOVED]

[SENSITIVE MATERIAL REMOVED]

[SENSITIVE MATERIAL REMOVED]

Appendix 14: Ethical approval letter 2 from the London Queen Square Research Ethics Committee

[SENSITIVE MATERIAL REMOVED]

[SENSITIVE MATERIAL REMOVED]

[SENSITIVE MATERIAL REMOVED]

[SENSITIVE MATERIAL REMOVED]

Appendix 15: Funding approval letter from Tourettes Action

[SENSITIVE MATERIAL REMOVED]

Appendix 16: Example plan for Session 2 CBIT child groups (Dr T.M., Tourette Syndrome Clinic)

[SENSITIVE MATERIAL REMOVED]

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Appendix 17: Example plan for Session 2 psycho-educational child groups (Dr TM, Tourette Syndrome Clinic)

[SENSITIVE MATERIAL REMOVED]

[SENSITIVE MATERIAL REMOVED]

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[SENSITIVE MATERIAL REMOVED]

Appendix 18: Examples of competing responses for different tics (Verdellen et al., 2011b)

[COPYRIGHTED MATERIAL REMOVED – See Verdellen et al., 2011b for further information]

Appendix 19: Outcome measures administered at pre- and post-assessment (including those for the wider study)

Outcome measures administered at pre-assessment	Outcome measures administered at post-assessment
Parents consent form and child assent form *If not yet completed, parents were given a pack of parent questionnaires to complete at their leisure during the assessment (SNAP-IV, parent CHOCI-R, SDQ, 4 items on rage attacks)*	Post-assessment questions asked from demographics questionnaire *If not yet completed, parents were given a pack of parent questionnaires to complete at their leisure during the assessment (SNAP-IV, SDQ, 4 items on rage attacks)*
GTS-QoL	GTS-QoL
TS visual analogue scale	TS visual analogue scale
NIH test: DCST (*using computer*)	NIH test: DCST (*using computer*)
NIH test: Flanker (*using computer*)	NIH test: Flanker (*using computer*)
NIH test: Motor dexterity (*using computer and 9-hole pegboard*)	NIH test: Motor dexterity (*using computer and 9-hole pegboard*)
PEDS-QL	PEDS-QL
PUTS	PUTS
WISC-IV (7 subtest short-form)	
15 minutes tic observation *(seated watching The Simpsons on laptop)*	15 minutes tic observation *(seated watching The Simpsons on laptop)*
5 minutes tic suppression *(seated watching The Simpsons on laptop)*	5 minutes tic suppression *(seated watching The Simpsons on laptop)*
YGTSS interview with child and parents	YGTSS interview with child and parents
Child CHOCI-R	

Notes. Outcome measures for each time point are listed in the order in which they were administered during testing. SNAP-IV is a measure of attentional difficulties. CHOCI-R = Children's Obsessional Compulsive Inventory - Revised; SDQ = Strengths and Difficulties Questionnaire; GTS-QoL = Gilles de la Tourette Syndrome Quality of Life scale; DCST = Dimensional Cart Sort Test; NIH = National Institutes of Health; *PEDS-QL = The Paediatric Quality of Life inventory; PUTS = Premonitory Urge for Tics Scale; WISC-IV = Wechsler Intelligence Scales for Children; YGTSS = Yale Global Tic Severity Scale*

Appendix 20: Pre-assessment protocol

Assessment Protocol Time 1 Assessment

Beforehand

- Tell other K/R where you will be and time of visit.
- Take contact details for TM and K/R
- Agree a time after which you will speak.
- Input demographic information into empty Excel file and save on F drive under ppt number
- Double check age and date of birth for eligibility
- Set up login for that child on the assessment centre and add password and login to the spreadsheet
- Make sure we know if they've had a WISC-IV already and input data if so
- Book travel. Bring address and contact details.

Equipment List (bold = things to replace each time)

- **ID badge**
- **Tickets and travel information**
- **Address of family; name, date of birth and age of child**
- **Participant number for child**
- Laptop + power cable
- Three-way plug adaptor
- Monitor, connector cable + power cable
- Keyboard, mouse, speakers
- Encrypted memory stick
- Pegboard, pegs and spares
- WISC-IV Blocks, stimulus book, scoring manual
- Stopwatch
- Pencil without rubber x 2
- **Little rubber man reward**
- **Bluetak**
- Internet dongle?
- DVD for watching during obs
- Full assessment protocol
- **Visual timetable**
- **Questionnaire pack for right age range (i.e. 13/ under 13)**:
    - *Participant assent form*
    - *YGTSS form*
    - *Young person CHOCI-R*
    - *WISC-IV record form response booklet*
    - *Tourette Syndrome VAS*
    - *PUTs*
    - *PEDs-QL (version different if aged 13)*
    - *GTS-QOL (version different if aged 13)*
- **Spare parent questionnaire pack in case they've lost theirs**:
    - *Parent consent form*
    - *Parent CHOCI-R*
    - *Rage attacks questionnaire*
    - *SNAP-IV 26*
    - *SDQ*

Equipment for Scoring

- Ruler, WISC-IV scoring templates, Clicker counter

Introduction
Introductions
Outline of assessment – Visual timetable
Reminder not to disclose group allocation if possible
Remind about GP letter if not already mentioned
Collect parent questionnaires and consent forms
Request internet password (2 mins)

Consent
Discuss study with parent and collect their written consent
Discuss study with child and collect their written assent (5 mins)

Initial questionnaires

1. GTS-QoL (NB: different questionnaire if age 13) (5 mins)

2. TS Visual Analogue Scale (5 mins)

Allow them to complete these on paper while we set up the computer equipment etc.

Unlock Encrypted file F by clicking on key icon.
Highlight file F and click mount. Usual password.

Right click desktop -> Screen resolution
Set up:
Display – 2. Acer (laptop)
Resolution – 1440 x 900
Orientation – Landscape
Multiple displays – Extend these displays

Open Excel spreadsheet for that participant number.

Setup three dongle or internet connection depending which is being used.
If dongle:
Plug in dongle and double click 3 icon.

If nesc turn on wifi on laptop – press fn then f3 (on laptop keyboard)

Go to Internet Explorer and Favourites – Choose TSGroupStudy

Neuropsychological Measures

3. Dimensional Card Sort (4 mins)
Only index finger

4. Flanker Inhibitory Control (3 mins)
Only index finger

5. Motor Dexterity task (Pegboard) (4 mins)

Test dominant hand first.
Position board horizontally with round container next to hand being tested, use bluetak to pin down. Put out spare pegs.
Demonstrate task
Practice and test trials for each hand
Other hand to be kept by side

Lay hand on table until told to go. 3-2-1 go…

Start the stop-watch as soon as the person touches the first peg
Stop the stopwatch as soon as the last peg hits the container.
[Record time with milliseconds for dominant and non-dominant hand)
Reposition the unit so round container is next to non-dominant hand. Repeat test.

6. PEDs-QL (NB: different questionnaire if age 13) (5 mins)

Enter data directly onto computer if possible as they complete the paper form
Can put equipment away at this stage if necessary or convenient

7. PUTS (5 mins)

Enter data directly onto computer if possible as they complete the paper form

8. WISC – 7 subtests (40 mins)

Block design, similarities, digit span, coding, vocabulary, matrix reasoning and symbol search

If a full-scale IQ from the WISC-IV is already available from previous testing and has been completed within the last 12 months, use this and skip WISC-IV testing.

9. Direct Obs while watching video (20 mins)

Set up video and camera
"Now we're going to film you, just to get a bit of a sense of what you're like. I'll put this video on so you can have something to watch and don't worry about the camera. It can be a bit of a break for you as well."
Encourage to have a drink, snack or use the toilet before starting.
Remind to stay seated so can see them on the camera.
Say **"Simpsons 1"**.
Start stop watch.

At 15 minute, say **"stop"**.

Label video Ppt number and assessment date and NS (non-supp) or TS (tic suppression)

"Now I'd like you to watch for another 5 minutes, but this time try your best to hold your tics in as much as you can for 5 minutes. After that I'm going to give you this stretchy man as a reward."
Then say **"Simpsons 2".** Start stopwatch.
Say **"stop"** after 5 minutes and stop video.
Label second video (see above)

Give stretchy man.

While child completes these measures, check their questionnaire filled in on paper for any missing items or unclear responses.
Score WISC and Visual Analogue Scale.

10. YGTSS (30 mins)

Ask permission from parent and child to video the interview for inter-rater reliability.
"Now I'd like to ask you both a bit more about the tics name has had in the last week."
Make sure they understand about:

- Sound tics
- Movement tics (can affect any part of the body, can give examples if necessary)
- Complex tics (e.g. Sometimes might have several that happen in a sequence)
- Tic signal – Urge and feeling better afterwards

Do initial background questions. Then:
"I'll start by asking you about your movement tics. In the last week have you, or have other people noticed any eye blinking tics?"
Go through list.

Coding notes
Complex – if includes a series of muscle groups or appears purposeful (e.g. motor hand through hair/obscene gestures or touching things; or vocal, saying a word).
Ver = verify (i.e. see in room).
Try to add e.gs. for the scores given where possible.
Ask initial questions about age of onset etc (but then for individual tics just ask about **current**)
Point out things you think are tics and check if they are (do they get tic signal? Is it unpleasant? How do you feel after the tic? Does it happen in different places?)
Make sure to differentiate between hyperactivity and tics, or OCD and tics.

"Now let's move on to your sound tics. Again, just thinking about the last week, have you, or have other people noticed any coughing tics?"
All specific e.g.s

"Now I've just got some more general questions about your tics".

FREQUENCY

- **"How often did your tics happen during the last week?"**

Follow up questions

  - Do you have at least one motor tic every day?
  - How about every hour, when awake on average?
  - How about every five minutes?
  - Do they occur in different places?
  - What's the longest time you've gone without ticing in the last week?

Look out for
If the reported frequency varies from what you observe ask about the discrepancy
It is not uncommon to tic more/less during discussion of tics

INTENSITY

"How forceful or strong are your tics?"

- Do they feel like they are bursting out of you really powerfully?
- How noticeable are your tics because of their intensity?
- You can ask how much others notice the tics (aside from family members and adults who know the child well)
- Use your own observations
- How exaggerated are the tics? Do they turn heads in public?
- Does it lead to pain/aches or injuries?
- Do you get scared of the tics? Would you turn your head? Higher scores then! If you doubt if someone coughs because of tics or because of having a cold, score lower.

Additional points
Noticeability is due to INTENSITY or STRENGTH, not frequency or complexity!

COMPLEXITY

How involved or orchestrated are the tics? – for us to code but ask more questions if necessary to clarify.

Additional points:
Usually rated based on observations and symptom checklist
If a complex tics includes both phonic and motor decide which is more dominant – do not rate twice.

Follow up questions:
If necessary ask about how hard they are to camouflage/how much they stand out due for:

- Duration
- Bizarre or obscene character
- Inappropriateness
- Unusual nature

INTERFERENCE

"How do tics get in the way when you're trying to do things? Like speaking or playing or doing things at school or at home?"

Additional points:
The key is the extent to which tics disrupt planned actions or speech
First establish if tics do interfere, then rate the extent
Use observations

IMPAIRMENT

"How much do tics affect your life? Are the tics stopping you from doing anything? Are you still able to feel good about all the great things you do?"

Queries
How do tic affect your:

- Self-esteem/mood ("feeling good about yourself and the things you can do/you've achieved")
- Enjoyment of things
- School, grades
- Relationships with friends, family
- Social acceptance, involvement, avoidance

Additional points
Impairment rated as a single item (not specified for motor/vocal, but rating the whole tic package concurrently)
0-50 scale

11. Child CHOCCI (15 mins)
Enter data directly onto computer if possible as they complete the paper form

12. Check over any items which were missing from the child questionnaires completed on paper

13. Check parent questionnaires (check over any missing items)

Save spreadsheet
Dismount the F drive

Extra info

To Log in to Assessment centre if necessary:
www.assessmentcenter.net in internet explorer

Appendix 21: Post-assessment protocol

Assessment Protocol Time 2 Assessment

Beforehand

- Tell K/R where you will be and time of visit.
- Contact details
- Agree a time after which you will speak.
- **Copy demographic information into new empty Excel file from time 1 spreadsheet and change assessment date. Save on F drive under ppt number and make clear T2.**
- **Set up new login for that child on the assessment centre and add password and login to the spreadsheet**
- Book travel. Bring address and contact details and give this information to K/R

Equipment List (bold = things to replace each time)

- ☐ **ID badge**
- ☐ **Tickets and travel information**
- ☐ **Address of family; name, date of birth and age of child**
- ☐ **Participant number for child**
- ☐ Laptop + power cable
- ☐ **Demographic info sheet**
- ☐ Three-way plug adaptor
- ☐ Monitor, connector cable + power cable
- ☐ Keyboard, mouse, speakers
- ☐ Pegboard, pegs and spares
- ☐ Stopwatch
- ☐ Pencil without rubber x 2
- ☐ **Little slinky reward**
- ☐ **Bluetak**
- ☐ Internet dongle
- ☐ DVD for watching during obs – **new disk**
- ☐ Full assessment protocol – **T2**
- ☐ **Questionnaire pack for right age range (i.e. 13/ under 13 based on age they were at first assessment)**:
    - *YGTSS form*
    - *Tourette Syndrome VAS*
    - *PUTs*
    - *PEDs-QL (version different if aged 13)*
    - *GTS-QOL (version different if aged 13)*
- ☐ **Parent questionnaire pack**:
    - *Rage attacks questionnaire*
    - *SNAP-IV 26*
    - *SDQ*

Equipment for Scoring

- ☐ Ruler, Clicker counter

Introduction
Outline of assessment – show visual timetable
Reminder not to disclose group allocation if possible
Give parent questionnaires
Request internet password

Go over T2 questions from demographic sheet: (2 mins)

- Since pre-assessment have there been any changes in medication?
- Since pre-assessment have there been any significant or stressful life events?
- Contact details still correct?
- Would they like to be contacted with regards to the findings of the study?
- Ok to possibly be contacted in 1 year for long-term follow-up?

Initial questionnaires

14. GTS-QoL (NB: different questionnaire if age 13) (5 mins)

15. TS Visual Analogue Scale (5 mins)

Allow them to complete these on paper while we set up the computer equipment etc.
Monitor to the left of the laptop

Unlock Encrypted file F by clicking on key icon.
Highlight file F and click mount. Usual password.

Right click desktop -> Screen resolution
Set up:
Display – 2. Acer (laptop)
Resolution – 1440 x 900
Orientation – Landscape
Multiple displays – Extend these displays

Open Excel spreadsheet for that participant number.

Setup three dongle or internet connection depending which is being used.
If dongle:
Plug in dongle and double click 3 icon.

If nec turn on wifi on laptop – press fn then f3 (on laptop keyboard)

Go to Internet Explorer and Favourites – Choose TSGroupStudy

Neuropsychological Measures

16. Dimensional Card Sort (4 mins)
Only index finger

17. Flanker Inhibitory Control (3 mins)
Only index finger

18. Motor Dexterity task (Pegboard) (4 mins)
Test dominant hand first.
Position board horizontally with round container next to hand being tested, use bluetak to pin down
Demonstrate task
Practice and test trials for each hand
Other hand to be kept by side

Lay hand on table until told to go. 3-2-1 go…

Start the stop-watch as soon as the person touches the first peg
Stop the stopwatch as soon as the last peg hits the container.
[Record time with milliseconds for dominant and non-dominant hand)
Reposition the unit so round container is next to non-dominant hand. Repeat test.

19. PEDs-QL (NB: different questionnaire if age 13) (5 mins)
Enter data directly onto computer if possible as they complete the paper form
Can put equipment away at this stage if necessary or convenient

20. PUTS (5 mins)
Enter data directly onto computer if possible as they complete the paper form

21. Direct Obs while watching video (20 mins)
Set up video and camera – **Mr Lisa Goes to Washington – Season 3 episode 2.**
"Now we're going to film you, just to get a bit of a sense of what you're like. I'll put this video on so you can have something to watch and don't worry about the camera. It can be a bit of a break for you as well."
Encourage to have a drink, snack or use the toilet before starting.
Remind to stay seated so can see them on the camera.
Say **"Simpsons 1"**.
Start stop watch.

At 15 minute, say **"stop"**.
Label video Ppt number and assessment date and NS (non-supp) or TS (tic suppression)

"Now I'd like you to watch for another 5 minutes, but this time try your best to hold your tics in as much as you can for 5 minutes. After that I'm going to give you this slinky toy as a reward."
Then say **"Simpsons 2".** Start stopwatch.
Say **"stop"** after 5 minutes and stop video.
Label second video (see above)
Give slinky toy.

While child watches the video, check their questionnaire filled in on paper for any missing items or unclear responses. Score Visual Analogue scale

22. YGTSS **(30 mins)**

Ask permission from parent and child to video the interview for inter-rater reliability.

"Now I'd like to ask you both a bit more about the tics <u>name</u> has had in the last week."

Make sure they understand about:

- Sound tics
- Movement tics (can affect any part of the body, can give examples if necessary)
- Complex tics (e.g. Sometimes might have several that happen in a sequence)
- Tic signal – Urge and feeling better afterwards

Do initial background questions. Then:

"I'll start by asking you about your movement tics. In the last week have you, or have other people noticed any eye blinking tics?"

Go through list.

<u>Coding notes</u>

Complex – if includes a series of muscle groups or appears purposeful (e.g. motor hand through hair/obscene gestures or touching things; or vocal, saying a word).

Ver = verify (i.e. see in room).

Try to add e.gs. for the scores given where possible.

Ask initial questions about age of onset etc (but then for individual tics just ask about **<u>current</u>**)

Point out things you think are tics and check if they are (do they get tic signal? Is it unpleasant? How do you feel after the tic? Does it happen in different places?)

Make sure to differentiate between hyperactivity and tics, or OCD and tics.

"Now let's move on to your sound tics. Again, just thinking about the last week, have you, or have other people noticed any coughing tics?"

All specific e.g.s

"Now I've just got some more general questions about your tics".

<u>FREQUENCY</u>

- **"How often did your tics happen during the last week?"**

Follow up questions

  - Do you have at least one motor tic every day?
  - How about every hour, when awake on average?
  - How about every five minutes?
  - Do they occur in different places?
  - What's the longest time you've gone without ticing in the last week?

<u>Look out for</u>

If the reported frequency varies from what you observe ask about the discrepancy

It is not uncommon to tic more/less during discussion of tics

<u>INTENSITY</u>

"How forceful or strong are your tics?"

  - Do they feel like they are bursting out of you really powerfully?
  - How noticeable are your tics because of their intensity?

- You can ask how much others notice the tics (aside from family members and adults who know the child well)
- Use your own observations
- How exaggerated are the tics? Do they turn heads in public?
- Does it lead to pain/aches or injuries?
- Do you get scared of the tics? Would you turn your head? Higher scores then! If you doubt if someone coughs because of tics or because of having a cold, score lower.

Additional points
Noticeability is due to INTENSITY or STRENGTH, not frequency or complexity!

COMPLEXITY
How involved or orchestrated are the tics? – for us to code but ask more questions if necessary to clarify.

Additional points:
Usually rated based on observations and symptom checklist
If a complex tics includes both phonic and motor decide which is more dominant – do not rate twice.

Follow up questions:
If necessary ask about how hard they are to camouflage/how much they stand out due for:

- Duration
- Bizarre or obscene character
- Inappropriateness
- Unusual nature

INTERFERENCE
"How do tics get in the way when you're trying to do things? Like speaking or playing or doing things at school or at home?"

Additional points:
The key is the extent to which tics disrupt planned actions or speech
First establish if tics do interfere, then rate the extent
Use observations

IMPAIRMENT
"How much do tics affect your life? Are the tics stopping you from doing anything? Are you still able to feel good about all the great things you do?"

Queries
How do tic affect your:

- Self-esteem/mood ("feeling good about yourself and the things you can do/you've achieved")
- Enjoyment of things
- School, grades
- Relationships with friends, family
- Social acceptance, involvement, avoidance

Additional points

Impairment rated as a single item (not specified for motor/vocal, but rating the whole tic package concurrently)
0-50 scale

23. Check over any items which were missing from the child questionnaires completed on paper

24. Collect and check parent questionnaires (check over any missing items)

Save spreadsheet
Dismount the F drive

Extra info

To Log in to Assessment centre if necessary:
www.assessmentcenter.net in internet explorer

Appendix 22: Demographic questionnaire

NHS Foundation Trust

Identification Number_____

Date _____

DEMOGRAPHIC INFORMATION
Parents: Initial telephone conversation

Hi this is name from calling about the Tourette Syndrome Group Study [if appropriate: returning you call]. Is now a good time to talk?

[If returning call] Thank you for contacting me and for your interest in the study. I'm calling today to see if you have any questions about the groups and the study. If you're still interested I'd also like to gather some general demographic information and ask some questions to make sure that your child meets the inclusion criteria to take part, that you would both be available on the necessary days and so on. Is it ok for us to go ahead with that now?

[If first contact from us] I'm calling today to check whether you received the letter and information sheets we sent in the post about our new study, in which we will be running groups for young people with Tourette Syndrome? If no – we can resend. If yes – Were you and your child interested in this? Would you like to ask any questions or like more information? If you are still interested, I'd just like to gather some general demographic information and ask some questions to make sure that your child meets the inclusion criteria to take part, that you would both be available on the necessary days and so on. Is it ok for us to go ahead with that now?

- Received information in the post? Yes No
- Read information sheets?
- Name of person you are speaking to and their relationship to child:
- Parent(s) and child able to attend groups at from 4.30pm for an hour or 90mins (depending on session) on either Wednesdays or Thursdays?

  Y / N

Child

- Name:
- Age:
- D.O.B:
- Gender:

- Ethnicity:
- What year in school is your child?
- Is English child's first language? Y / N
  - ➔ If not, what is their English ability and what other languages do they speak?
  - ➔ PREVIOUS INPUT: Has your child ever had any counselling, therapy or any other kind of treatment for their tics? *(What? When? Where? How many sessions?)*
  - ➔ CURRENT INPUT: Is your child currently having any counselling, therapy or any other kind of treatment for their tics? *(What? When? Where? How many sessions?)*
- How do you think they would cope with being in a group?

Has your child been diagnosed with:

Tourette's Syndrome ☐
Chronic Vocal Tic Disorder ☐
Chronic Motor Tic Disorder ☐
ASD ☐
OCD ☐
ADHD ☐
Learning Disability ☐
Other ☐

List of medications (current): [Name, dose, how long been taking]

Parents/Guardians

- Name (title, first and last names) of other parent/guardian (not spoken to):

- Ethnicity of parents:

- HOLLINGSHEAD SES:

1) Gender of parent(s) in household – [No need to ask if obvious from previous questions]:

Total SES score: _____

Score: ____

2) Marital status *(living together?)*:

Score: ____

3) Parent's/guardian's level of education (both):

Mother:

Father:

Score: ____

4) Parent's/guardian's occupation (both): *Currently working? What?*

Mother:

Father:

Score: ____

- Live in a house/flat? Council-rented? Privately rented/owned?

- Number of other people living in the house:

- Number of brothers/sisters:

Other details

- Contact number(s):
- Best time of day to contact:
- Contact address:
  - ➔ Estimate of travelling time if outside of London:
- Do you have high-speed Wi-Fi at home?

Eligible to take part? Y / N

Child interested in taking part? Y / N

[Check it has been discussed with child – Child info sheet read?]

Parent still interested in taking part? Y / N

[If no to any, please write down reasons why if known:]

When would be best to conduct the pre-assessment? (availability)

Re home visit appointment & what will happen next

Is there a quiet space and table available at home that we could use?

Please could they have their internet passwords available on the day?

We'll send them out the questionnaire packs in the post – They can complete them, or hang onto them and do them on the day we come, whichever is more convenient.

We'll pass their details on to the clinical team who will carry out the randomisation. They'll then be in touch about the groups.

Reminder of what will happen re randomisation and that we will not be involved. Therefore we ask them please to not mention when we meet the group to which they have been assigned.

Pre-assessment date arranged?

Thank you very much for speaking with me today. [Confirm meeting date arranged or say "I'll be in contact as soon as possible to arrange a meeting date with you to meet child and conduct the pre-group assessment].

General comments: *(Any concerns? Things for clinicians to keep in mind?)*

Post-Assessment follow-up **Date:**

- Since pre-assessment have there been any changes in medication?
- Since pre-assessment have there been any significant or stressful life events?
- Contact details still correct?
- Would they like to be contacted with regards to the findings of the study?
- Ok to possibly be contacted in 1 year for long-term follow-up?

General comments:

Appendix 23: Simpsons episode selection for direct tic observation measure

Children in both groups watched the same Simpsons episode at pre-assessment. At post-assessment, unlike Himle and colleagues (2006), a different episode was shown to ensure maximal engagement. Researchers felt this was important as different levels of stimulation, relaxation and other factors have been indicated to influence tic expression (Conelea & Woods, 2008). Researchers carefully reviewed episodes to ensure they were age-appropriate. Three potential new episodes for post-assessment were selected by the researchers, to be rated by a 14 year old girl (a cousin of one of the researchers) in terms of such factors as reported by Conelea and Woods (2008). The video most similar to the pre-assessment episode, in terms of these ratings, was chosen (see below).

A) "Homer Simpson, This is your Wife" – Season 17, Episode 15 [ORIGINAL VIDEO, PRE-ASSESSMENT]

http://www.wtsof.tv/watch/S17E15-homer-simpson-this-is-your-wife

1. How stressful/ anxiety provoking was this episode?

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

2. How boring was this episode?

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

3. How relaxing was this episode?

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

4. How stimulating was this episode? (i.e. funny/exciting?)

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

5. How upsetting or sad was this episode?

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

6. How frightening was this episode?

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

B) "Mr Lisa Goes to Washington" – Season 3, episode 2 [NEW VIDEO 1]

http://www.wtsof.tv/watch/S3E2-mr-lisa-goes-to-washington

1. **How stressful/ anxiety provoking was this episode?**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

2. **How boring was this episode?**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

3. **How relaxing was this episode?**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

4. **How stimulating was this episode? (i.e. funny/exciting?)**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

It wasn't really funny or exciting but it had interesting themes?

5. **How upsetting or sad was this episode?**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

6. **How frightening was this episode?**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

C) "Lemon of Troy" – Season 6. Episode 24 [NEW VIDEO 2]

http://www.wtsof.tv/watch/S6E24-lemon-of-troy

1. **How stressful/ anxiety provoking was this episode?**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

2. **How boring was this episode?**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

3. **How relaxing was this episode?**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

4. **How stimulating was this episode? (i.e. funny/exciting?)**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

5. **How upsetting or sad was this episode?**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

6. **How frightening was this episode?**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

D) "Bart Vs Australia" – Season 6, Episode 16 [NEW VIDEO 3]

http://www.wtsof.tv/watch/S6E16-bart-vs-australia

1. **How stressful/ anxiety provoking was this episode?**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

2. **How boring was this episode?**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

3. **How relaxing was this episode?**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

4. **How stimulating was this episode? (i.e. funny/exciting?)**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

5. **How upsetting or sad was this episode?**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

6. **How frightening was this episode?**

Not at all 0 – 1 – 2 – 3 – 4 – 5 – 6 – 7 – 8 – 9 – 10 Extremely?

	Stressful	Boring	Relaxing	Stimulating	Sad	Frightening	Total point difference to original video score
Original video	**1**	**4**	**7**	**5**	**0**	**0**	N/A
New video 1	2	4	6	7	2	0	6
New video 2	3	6	4	6	0	2	10
New video 3	5	3	3	4	2	4	16

Appendix 24: Tic observation scoring protocol

TS Group Study: Tic Counting Protocol

Some background information:

- In this study children are seen once before (**pre**) and once after (**post**) attending group therapy for tics.

- In the pre- and post-visits, children are filmed using the laptop Webcam whilst watching a Simpson's episode for **15 minutes** in order for us to count how many tics they display within that time (**non-suppression task - NS**). They are then asked to hold in their tics for **5 minutes** (**tic-suppression task - TS**) whilst watching the rest of the Simpsons episode. You will hear us say "Simpsons 1" at the beginning of a video to indicate that the task is non-suppression, and "Simpsons 2" to indicate that the task is tic-suppression.

- You will be asked to **count tics** (vocal and motor) that you observe within those time periods, following the directions outlined below and using a tic counter.

- You will **find the videos** for each participant on the **hard-drive** in the drawer below the printer in room ____ with the **yellow label "tic counting"**.

- The videos are separated into **"M" and "K" videos** – This relates to whether the videos were taken in **pre- or post**-group visits (these letters have been chosen at random to refer to a particular set of videos). This is to allow you to remain unaware of the time point to ensure fair scoring.

- Each person should have a total of 4 videos, 2 in "M" and 2 in "K" (at each pre- and post-visit each child will have done 1 x 15min NS and 1 x 5min TS).

- We would like you to score all of one participant's videos (M and K) before moving onto the next participant. We would like you to change the order for each new participant you score. For example, if for one participant you scored the A videos and then the Bs, for the next participant you should score the B videos and then the A. We have reminded you to do this by changing the order each time on the Excel data entry file (which will be explained below).

- Each participant has a semi-structured interview with parents and children in pre- and post-group visits. This is called the Yale Global Tic Severity Scale (**YGTSS**) and involves us noting down on a list all of the motor and vocal tics the child has had in the last week. We have written out a complete list of tics for each person ("tic list"). **You will just be rating these only, and no other tics that you see in the videos that are not on the list. You can find a list of tics for each person in the see-through folder on the shelf above my desk, between the TS Group Study folders.**

  DIRECTIONS

- Ensure you have a tic counter (there is one in K's top drawer), scrap paper, pen, stopwatch (can use ones on phones) and headphones (if possible/necessary).

- Get one of the hard-drives from the drawer and plug both of the hard-drive USB connectors into the computer. Go to My Computer and double click on McAfee then Start. Click login - Password is ___. Then go back to My Computer and open the Private (E:) file that has now appeared. Go to the **"TS Group Study Videos"** and then the **"Tic counting" file**.

- Open the Excel spreadsheet for tic counting, where you will enter the data. Start at the top and work your way down. 1 participant = 2 rows, 1 for M and 1 for K time points.

- Look at the first column and get the ID number of the participant you will score on the Excel sheet. Find their tic list (on the shelf above my desk, see-through folder, yellow label "tic counting") and look through the list of tics that came up for that person on both visits, so that you are aware of which tics you will be counting in the videos.

- Now go to the file "tic counting videos: first (or second, depending on participant number you're looking for) set of groups", and find their videos in the "M" file or "K" videos file for that participant, starting with whatever is first (working top to bottom) on the two rows in the Excel file for that person.

- Within the "M"/"K" file, look at the 15min NS video first. Have your tic counter ready. We would like you to **click on the counter every time you see any tic (motor or vocal) that is on the list for that person (DO NOT COUNT TICS THAT ARE NOT ON THE LIST).** Make sure you **do this for exactly 15mins and no more or less** ("time start/stop" in the table below i.e. 0m 40s / 15m 40s is to remind you this should be exact). Videos often go on for longer but shouldn't be scored for more than the allocated time. You can pause the video if you need to check the list at any point.

- If during this time there are moments when it is difficult to observe the participant fully (e.g. they turn away, put their head down, leave the room) you should time these with your stopwatch. It is probably easiest to pause the stopwatch and continue it the next time, so at the end of that task you have a total number of seconds in which the tics were not fully observable. If the video is paused that is fine, just continue when we say start again. [NB: This method of measuring tics is not perfect and we understand it's limitations i.e. difficulty in observing tics from the waist down due to filming on the table, children often fidgeting etc – Please only rate tics as unobservable/nonvisible if it is quite significant e.g. they turn around, they get up and walk away briefly, they bend down to the ground to get something, and so on]

- **Fill all information in on the table below the tic list for that participant (hard copy)** – start/stop time, total tics, total time tics were unobservable/nonvisible, comments and any other tics you saw that were not

on the list and so were not counted (write these down as you go along – you may need to pause the video).

So BASICALLY as you are watching the video make sure you:

- Write down the **start/stop time**
- **Count the number of tics** you see (only those on the tic list)
- Use your stopwatch to record **total time** tics/participant was **unobservable**
- **Write down any other tics you see that are not on the tic list** (don't count them, just write down what they were i.e. head jerk, repetitive "uh" vocal tic, etc)
- Write down any **comments**

NB: *Make sure you write down the results for M/K in the correct columns on the tic list sheet! This is a common mistake that is easy to make.*

- **Excel data entry:** Go to the database and the row for that participant and the time point (remember **M and K** time points mean that each person has two rows). Write the number of tics counted (e.g. 20) and the total time the tics were unobservable/nonvisible (as measured using your stopwatch - SECONDS). The pale green columns are calculated automatically.

- Now go back to the videos and do the same for the 5min TS video (i.e. count the tics using the tic counter, use the stopwatch to monitor time tics were not fully observable).

- Record this data on the tic list sheet and in the Excel data entry file - in the columns to the right on the same row of the Excel sheet for that participant.

- You have now entered all of the data for the "M" or "K" time point for that participant, and need to do the same for the other time point ("M" or "K" video files) that have not yet been rated for that participant.

- Enter these results on the row below for that participant.

- DONE! Onto the next participant/row.

COMMON DIFFICULTIES AND A FEW THINGS TO REMEMBER!..

Please be very careful to enter the correct data in the 2 rows for each person, relating to the "M" and "K" time points.

Make sure that you are only counting tics for EXACTLY 15 or 5 minutes, as videos often go on slightly longer.

On very few occasions we may have forgotten to press stop on the video and the 15min NS and 5min TS tasks may all be on one video – Just listen out for "Simpsons 1/2" to indicate the start of timing, and "stop" (or something like saying we're finished now) to indicate the end of timing.

Sometimes us saying "Simpsons 1" or "Simpsons 2" at the beginning of each video may not be audible. In this case simply start at an appropriate point, such as when the examiner walks away, and then count for the allotted time.

Do not count general fidgetiness as tics (e.g. playing with face, hair, objects etc). If something really goes wrong you can re-do the video again, but this will take much more time so we'll avoid that if we can. PLEASE COME AND FIND US IF THERE ARE ANY PROBLEMS OR YOU HAVE ANY QUESTIONS – This data is very important so we would rather you asked us more than less questions! There is also a comments section on the right in the Excel file in which you can record any difficulties or issues, but these should always be discussed with us first ideally.

NB: An error/danger message often comes up when using the hard-drive. Rather than continually pressing close just drag it to the bottom of the screen and continue with your work.

What if…?

➔ **I am unsure whether or not a vocalisation/movement is a tic or not?** You will have seen the YGTSS so will know some tics to look out for, but if you are not sure just make sure you are consistent. If you have counted something as a tic before then the best thing to do is keep counting it as a tic throughout, and vice versa if you have not.

➔ **The child has very complex tics and it is hard to tell what is one tic e.g. long juddering tic with eye twitching and vocal tics at the same time.** Generally count complex tics ONCE (e.g. a sequence of orchestrated tics – there may be breaks of only a second between bouts of complex tics but try to spot these and count separately. <u>We will go through some examples beforehand.</u>

If someone has tics in which they say lots of words all at once without a break count these as one tic.

If someone says sentences then count each sentence as one tic.

➔ **I can't hear quiet vocalisations well on the video?** Sometimes quieter sound tics are not quite audible on the videos and this is a limitation of this method. Unless you can see mouth movements that clearly indicate a sound tic, it is not possible to improve the sound quality to capture less audible vocal tics.

**** THANK YOU FOR YOUR HELP! ****
Please let us know if you have any questions – One of us should usually be around or you are welcome to phone us.

Appendix 25: Tic observations: Tic list and scoring sheet

Tic list and scoring

Ppt number:

MOTOR TICS

VOCAL TICS

LESS LIKELY TO SEE ON VIDEO

	15 mins				5 mins			
K	Start time	Stop time	Tic count total	Secs non-visible	Start time	Stop time	Tic count total	Secs non-visible
	Comments							
M								
	Comments							
Clear tics seen but not listed and so not counted								

Appendix 26: General details of the tic observation scoring process

For each child, raters were given a detailed protocol (see Appendix 26) and “tic list and scoring” sheet (see Appendix 27). This was a list pre-prepared by the researchers (as in Himle et al., 2006), in which all motor and phonic tics that had previously been reported in the YGTSS interview (combining both time points) were listed. This was essential in terms of scoring consistency and inter-rater reliability, as tics and non-tic behaviours were often very hard to differentiate. Raters were then asked to watch the four videos for that child (tic observation and suppression conditions; pre- and post-treatment) and record from each video the number of tics they saw. This followed an event frequency method, as opposed to partial-interval coding in which segments of a video would be observed. Both approaches have been shown to be appropriate and highly correlated (Himle et al., 2006). Raters were provided with a mechanical tic counter. The number of tics observed (from the tic list only) was entered into a pre-prepared Excel spreadsheet along with the number of seconds the child was not adequately visible (e.g. turned around, got up, covered their face). A total tics per minute index score was then automatically calculated for each video.

As tic lists were often lengthy, raters were asked to score one child at a time in order to get a good impression of the child and their tic expression. Raters were blind to treatment condition and time point. Anonymous study ID numbers were used to identify each child. Time point was labelled as “M” or “K”. Raters scored half of the videos each. Prior to scoring, raters were fully trained by the researchers, using the same practice videos and engaging in discussions when any discrepancies arose.

Appendix 27: Invitation letter

NHS

NHS Foundation Trust

[Name]
[Address]

[Date]

Dear [Name],

I am writing to invite you and your child to participate in a study which my colleagues and I are carrying out at We are contacting you because your child has been seen at our clinic in relation to their Tourette Syndrome or chronic tic disorder.

Please find enclosed an information sheet explaining the study. It outlines our reasons for conducting the study and what would be asked of you and your child should you choose to participate. There is also a children's version of the information sheet included which you can discuss with your child should you wish.

If you and your child are interested in taking part, or would like to know more, please contact one of my colleagues (Katie Edwards, University College London, tel: , or , Royal Holloway University of London, tel:). Further details are provided on the information sheets attached.

If we do not hear from you, we will contact you by telephone in about two weeks to check you have received this information and to discuss any queries you may have.

Many thanks for taking the time to read the enclosed information.

Yours sincerely,

Dr
Consultant Clinical Psychologist

Appendix 28: Parent / carers information sheet

NHS Foundation Trust

Information about the project

Project title: Randomised pilot study evaluating two group therapies for Tourette Syndrome

We work at the Tourette syndrome Clinic at We would like to invite you and your child to take part in a research study. Before you decide if you would like to take part, it is important for you to understand why the research is being done and what it will involve. Please read through the following information carefully and discuss it with others if you wish. Ask us if there is anything that is not clear or if you would like more information. Take your time to decide whether or not you wish to take part. This would involve attending group therapy as well as completing assessments before and after so that we can evaluate whether the group has been effective.

What is the purpose of the study?

This study is interested in group based psychological therapy for Tourette Syndrome and other chronic tic disorders. As you probably know, the symptoms of these disorders, including tics themselves, can impact greatly on the quality of life of those who experience them. This can be either directly, in terms of physical discomfort associated with the tics themselves, or indirectly, as a result of factors such as the reactions of other people or by making it difficult to concentrate on school work. It is therefore very important that we find effective means of treating the symptoms.

Currently, treatments are usually delivered individually to each child. This study aims to investigate whether delivering therapies to groups of children could be equally, or more, effective. It is possible the children will benefit from the chance to meet other children experiencing similar difficulties to their own. In the long run, if such treatments are shown to be effective, it could increase the number of treatment options available to families and potentially also reduce waiting times for therapy.

Why have my child and I been asked to help?

We are asking children, aged 9-13 years, who have been seen previously at or who have recently been referred to the clinic, if they are interested in participating.

Do I have to take part?

No. Taking part in this study is entirely voluntary. If you decide not to take part in this study, you do not have to give a reason, no one will be upset and the standard of care your child receives will not be affected. If you do decide to take part, you can still withdraw at any time, without giving a reason, even if your child has started attending the group.

Group sessions

To participate in the group based psychological therapy it will be necessary for your child to attend the clinic at for 8 weekly sessions. These will run either from September to October or November to December 2013. The groups will run from 4:30pm on a week night. The first two sessions will be 90 minutes long and subsequent sessions will last for an hour.

The groups will be a chance for your child to make friends, meet other children with a tic disorder and learn about how others cope with their symptoms. In the groups the children will participate in various games and puzzles and in the final session there will be a party.

The exact content of the group will depend on the group your child is assigned to (see below). One set of groups will be based on Habit Reversal Therapy, in which your child would be taught particular techniques aimed at helping control their tics. The second set of groups will focus on learning about topics such as school and bullying, self-esteem and dealing with difficulties in relation to anger, attention and impulsivity.

Following each session, your child will be set a small 'homework' task. This will be something to practice or do with your child during the week.

During sessions number three to six, parents and carers will also be invited to attend a 4-session parent group which will run at the same time as your child's group. This will include learning about Tourette syndrome and developing strategies to help your child cope with their tics. These sessions are intended to complement the work that your child will be doing in their group.

In which group would my child participate?

As the study will compare two group based therapies using different theoretical approaches, children will participate in either one program or the other. No scientific research has previously investigated the effectiveness of either group and therefore we do not know which may be more beneficial. In order to make it a fair test we need to allocate children to the groups randomly. Therefore, once you decide to take part in the study we would enter your child's participant number into a computer programme, which would randomly assign them to one of the two groups. We would then let you know which group had been assigned and provide you with more information about the days when the group will take place.

Apart from attending the groups, what else will my child and I be asked to do if we take part?

We would initially send you some questionnaires for you, the parents or guardians, to complete at home. We would then telephone you to arrange an initial assessment. We could visit you at home or meet at depending on which is most convenient for you.

During the first visit, we would complete some puzzles, tasks and questionnaires with your child and expect that these would take about 2 hours and 50 minutes, including rest breaks. During this time, they would also spend 20 minutes watching a DVD while we video them. For the final five minutes of this section, we would ask your child to reduce their tics as much as they are able.

Following this initial assessment, your child would be invited to participate in a group therapy program at . On completion of the group, we would arrange two further assessments to assess any change that may have occurred. These visits would be expected to last about 1 ½ hours, again including rest breaks. The first would take place within a month of finishing the group and the second 1 year later. We would also ask you as parents and carers to complete some additional questionnaires prior to each of these assessments.

Is there anything to be worried about if my child and I take part?

There are no specific risks from taking part in the study as your child's treatment will not be changed by participating the study in any way. If your child gets tired when we are doing the tasks and puzzles then they will be able to take breaks.

It is possible that thinking about their life and the effect of having a chronic tic disorder could be upsetting for your child. If the questionnaires do cause any distress, I would ask that you let us know so that we can offer support and think about what further help is needed.

Will my child's tics increase?

It is possible you may be concerned about your child being exposed to other children with different tics in case your child's tics become worse or they adopt new tics. While this is

possible in the short-term, clinical experience has tended to show that this is not a lasting effect. Nonetheless, should you have any concerns at all during the study you should discuss these with us and you would be entirely free to withdraw from the study at any time should you wish.

Will taking part help my child?
These particular groups have never been tested for children with tic disorders and therefore we do not know whether your child will experience any benefit from their participation. The groups have been designed, based on what is currently known, to help children with chronic tic disorders. We therefore hope that your child might experience some benefit in some areas such as their quality of life or the intensity of their tics.

How will the information help people?
When the study has finished we will write to you to let you know what we found out about the groups. We hope this study will help us find out if group therapies are useful for children with tic disorders.

Will my child's usual treatment be affected by taking part?
If your child is currently receiving treatment at they would continue to be seen as a patient here throughout the study. Any school liaison work, or medication, would continue as normal and be unaffected by participation. If you choose to take part, the only difference would be that your child would not be able to participate in any individual therapy during the study. If your child has been offered this, and you would prefer this to a group based intervention, it would be best not to participate in this study. Should it be felt during the course of the study that your child may benefit from individual therapy, you would be able to withdraw from the study and receive this treatment as appropriate.

Who will know that my child and I are taking part in the study?
All information that is collected about your child during the course of the study will be kept strictly confidential. We would keep your and your child's name, address and results from the puzzles and questionnaires secret. We would also keep all paperwork in a safe place. After we have watched the videos of your child, in order to count their tics, the videos will be permanently deleted. We would write about the study but no names would be used or any information that would show it was your child. If you agreed then we would write to your GP to let them know you are taking part in the study.

What will happen to the results of the study?
The results will not be known until the last groups are run and the data is collected for everyone taking part in the study. We hope to complete the short-term outcome of taking part in the groups by early 2014. Following this, we would hope to meet again one year later in order to see whether any effects of the groups are maintained. The results may appear in professional publications and meetings and as part of a doctoral university assignment, but neither you, nor your child, would be recognisable from any transcription. We will also write to you at the end of the study with a brief summary of what we found out. We hope to hold a general feedback session once the study is complete, which you will be invited to.

Who has organised and approved the research?
All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and approved by the London Queen Square Research Ethics Committee. Their contact details are provided below. This research has also been reviewed and approved by ethics committees at Royal Holloway, University of London and University College London. The research is being sponsored by

Who is funding the research?
Funding for the study has been provided from three sources. These are Royal Holloway, University College London and the Tourette Action, UK (the National Charity for Tourette syndrome).

What if something goes wrong?
This study is indemnified under the Clinical Negligence Scheme for NHS Trusts, which provides cover for negligent harm. If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions. If you remain unhappy and wish to complain formally, you can do this via the Patient Advice and Liaison Service at (You can ring them on or email them on).

What do I do now?
Thank you for reading this information. If you and your child are interested in taking part in this study, please contact (Tel: or Katie Edwards (Tel:) to hear more. If we do not hear from you, we will contact you by phone in about two weeks to answer any questions you may have and to see if you are interested in taking part.

Who do I speak to if I have further questions or worries?

Contact: Katie Edwards, Trainee Clinical Psychologist
Trainee Clinical Psychologist

Address: TS Group Study

Email:

Tel: Katie Edwards:
Rachel Yates:

Supervised by: Dr , Consultant Clinical Psychologist,

Contact details for London Queen Square Research Ethics Committee:
HRA Head Office, Skipton House, 80 London Road, London, SE1 6LH
Telephone: 020 7972 2584

Appendix 29: Child information sheet

Participant information sheet
for children and young people
Version 3 10/05/2013

NHS Foundation Trust

Information about the project

Project title: Randomised pilot study evaluating two group therapies for Tourette Syndrome

We work at Hospital. We are asking you and your parents to take part in a project. This leaflet will tell you about the project. We hope you can read about the project with someone in your family. Please ask us if you have any questions. Take your time to decide whether or not you want to take part.

What is this project and why are we doing it?
This study is interested in whether new therapies can help with difficulties experienced by children with tic disorders. The treatments involve weekly sessions at the hospital with psychologists (talking doctors) along with a group of other children who also have tic disorders similar to you. During 8 sessions, the psychologists will help teach you all some things you can do to manage your challenges. We would like to find out if coming to these groups helps you in your life or makes your difficulties easier to cope with.

Why have I been asked to take part?
We are asking all children who have visited Hospital for help with a tic disorder to take part in the study.

Do I have to take part?
No, you do not have to take part. If you decide not to take part in this study, you do not have to give a reason and no one will be upset. You can change your mind at any time. You can stop being in the study even if you said yes at the beginning or if you have already started attending the groups.

Will taking part help me?
We don't know for sure if the groups will help you. We hope that it will help you to cope with some aspects of your tic disorder. We will evaluate whether it has helped you to reduce your tics or whether it has helped you in other areas such as your satisfaction with your life in general or your ability on certain thinking tasks. Afterwards, we would let you know if the groups helped you in terms of any of the areas we have evaluated.

As an additional reward for taking part, we will enter all children or young people who have attended 6 or more of the group sessions into a prize draw to win £50 of gift vouchers.

Once the study is finished we will invite you and your family to a feedback session where we will explain the results of the study and what we have learnt.

What will I be asked to do if I take part?

Meeting 1



First we would arrange a meeting with you and your parents or carers at home or at Hospital. At this meeting:

- One of us would spend about 2 hours 50 minutes with you doing puzzles and asking you some questions.
- We will ask you to do a selection of different things and hope you will find them interesting.
- During the visit we would ask you to watch a DVD for 20 minutes and make a film of you as you watch it. For five minutes we would also ask you to try to tic as little as you can.
- You would be able to have short breaks if you feel tired or to stop if you want to.
- We would also ask your parents or carers some questions.

Coming to the groups

If you choose to take part you would come to 8 group therapy sessions at Hospital along with a small group of other children or young people with tic disorders. We will invite 12 children to each group. The groups will run in the afternoon, at 4:30pm on one evening a week. The first two sessions will last 1 ½ hours and the rest 1 hour. During each session there will be a variety of things to do and we hope you will find them interesting.

In each session we will also give you a small 'homework' task of something to do or practice at home with the help of your parent or carer.

During some of the sessions we will also invite your parent or carer to come to their own group to learn something about what you have done in the groups and how best to help you keep using the things you learn at home.

Meetings 2 & 3


When you have finished the 8 group sessions we would meet again twice more, either at your home or at the hospital. We would meet a few weeks after the groups and then again 1 year later.

At these visits we would do more games and puzzles and ask you and your parent or carer some more questions. This time the visit would be a bit shorter, probably lasting about 1 ½ hours.

Is there anything to be worried about if I take part?
When we do the games and puzzles you can take breaks if you get tired.
We will make the groups as fun as possible. If you are upset by taking part in the study, please speak to your parents about it. If you would like to speak to someone else, your parents know how to contact us and our address and phone number are at the end of this sheet. Your treatment at Hospital will not be changed by taking part.

Who will know I am taking part in the study?
We would keep your name, address and your results from the games and puzzles secret. We will write about the study but no names will be used. If you agreed then we would write to your doctor to let them know you are taking part.

What will happen to the results of the study?
The results will not be known until about September 2014. We hope to organise a time to tell everyone about the study soon after that, which you would be invited to.

Who do I speak to if I have further questions or worries?
Your parents also have information about this study. You can ask them questions. You can contact Katie Edwards or if you have any other questions.

Contact: Katie Edwards, Trainee Clinical Psychologist
, Trainee Clinical Psychologist

Address: TS Group Study

Email:

Tel: Katie Edwards:

Supervised by: Dr ', Consultant Clinical Psychologist,

Appendix 30: Parental consent form

NHS

NHS Foundation Trust

Identification Number_____

CONSENT FORM FOR PARENTS/ GUARDIANS

Title of Project: Randomised pilot study evaluating two group therapies for Tourette Syndrome

Names of Researchers: Trainee Clinical Psychologist
Katie Edwards, Trainee Clinical Psychologist
Dr Consultant Clinical Psychologist
Dr ı, Consultant Child Psychiatrist

Version and date of the participant information sheet that the parent/carer has read:_____

Please initial the box after each statement.

1. I confirm that I have read and understood the information sheet for the above study. I have had the opportunity to consider the information, ask questions and have had these questions answered satisfactorily. ☐
2. I understand that my child's participation is voluntary and that I am free to withdraw at any time, without giving any reason, without medical care or legal rights being affected. ☐
3. I agree to my child being videoed for the purpose of the study. ☐
4. I understand that the videos will be encrypted and stored on password protected computers. They will be permanently erased once they have been viewed by the researchers, in line with data information governance and the law. ☐
5. I understand that while participating in the study my child will be unable to receive any individual psychological treatment, but that should they require this, we will be able to withdraw from the study at any time. ☐
6. I understand that sections of my child's medical notes may be looked at by the researchers where it is relevant to my taking part in the study. I give permission for these individuals to have access to my child's records. ☐
7. I agree to my child's GP being informed of their participation in the study. ☐
8. I agree to taking part in the above study. ☐

Name of Child_____

_____	_____	_____
Name of Parent or Guardian	Date	Signature
_____	_____	_____
Researcher	Date	Signature

Appendix 31: Child assent form

Participant assent form
Version 2 04.05.2013

Identification Number_____

NHS Foundation Trust

PARTICIPANT ASSENT FORM

Title of Project: Randomised pilot study evaluating two group therapies for Tourette Syndrome

Names of Researchers: Trainee Clinical Psychologist
Katie Edwards, Trainee Clinical Psychologist
Dr , Consultant Clinical Psychologist
Dr , Consultant Child Psychiatrist

Please circle YES or NO

Have you understood the information you were given?	YES	NO
Have you understood that we would video you as part of the study? The videos would be deleted once we have used them and will not be viewed by anyone except the researchers.	YES	NO
Have you been able to ask questions and had them answered?	YES	NO
Would you like to take part?	YES	NO
Do you understand that you can stop being involved in the study at any time you like?	YES	NO

_____	_____	_____
Name	Date	Signature
_____	_____	_____
Researcher	Date	Signature

Appendix 32: Example of visual timetable used in pre-assessments

PLAN FOR TODAY

- 3 forms 
- Computer games x 2 
- Peg game 
- 2 forms 
- Games and puzzles 
- Watching a Simpsons episode 
- Trying to hold in your tics for 5mins → Stretchy man REWARD! 
- Talking together with your parents/carers about your tics this week 
- 1 form 

THE END

THANK YOU! ☺

Appendix 33: Letter to GP

[GP Address]

NHS Foundation Trust

[Date]

Dear Dr [Name],

Patient details:

The above named patient has consented to participate in a research study at Hospital, which aims to investigate group work for children with Tourette Syndrome or chronic tic disorders. It is a randomised pilot study to evaluate the efficacy of two treatments: A tic-specific behavioural intervention versus a psycho-educational group.

If you would like any further information on the study, please do not hesitate to contact us (correspondence address: TS Group Study,

Yours sincerely,

Katie Edwards
Trainee Clinical Psychologist

Trainee Clinical Psychologist

Supervised by: Dr , Consultant Clinical Psychologist and Paediatric Neuropsychologist

Appendix 34: Randomisation instructions

Thank you for agreeing to help us with the randomisation process for the Tourette Syndrome Group Study!!

Here are what we hope are some simple instructions on how to carry out the randomisation process.

1. Go to http://qminim.saghaei.net/index.php
2. Login: XXX; Password: XXX)
3. Go to the subjects tab
4. Click the green plus sign on the left hand side of the screen
5. For each new participant, add their gender and age and then click done. The program will then assign them to either the HRT or the Psych-Ed group.
6. If you add them in order of participant number in the study then the ID number assigned by the program should match our participant number. This may not always be possible, so if they are not matching, then please just keep a track of the numbers assigned by the programme so we know which participant is which.
7. Please do not ever click on the tab at the top that says new, as this will delete the study and we'll need to start from scratch.
8. We have created an Excel spreadsheet called "Randomisation spreadsheet" to help keep track of the group to which each participant has been assigned.

We hope this makes sense. If not, please let us know and we will do our best to help.

Many thanks.

Appendix 35: Further details of practical and ethical considerations

All participants were given a confidential study ID number, which was used on all electronic databases and assessment measures. The main electronic database that held study ID numbers in combination with patient-identifiable information was securely stored on a password-protected computer in a locked room at the hospital. All paper-based documents were stored in a locked room at the hospital, which was only accessed by the clinical team.

All parents and carers were requested to remain at home until testing was complete. Electronic data gathered during assessments was stored on laptops with study ID numbers only in a password-protected file within an encrypted hard-drive, before being transferred to the secure database at the hospital (or encrypted hard-drive for videos) and deleted from the laptop. The NIH online measures only identified participants by age and a unique code.

Dr TM provided supervision for both researchers throughout the study and was the main point of contact for any arising concerns or research issues. Information was also shared with the clinical team as appropriate. Any arising concerns were acted on in accordance with NHS and the hospital's policies and procedures.

Appendix 36: Further details on service user feedback (mother of a child with TS)

The mother said that the parent information sheet was highly informative and answered all of the questions she would have. She liked the question and answer format and did not find the amount of information provided off-putting, stating that it gave parents the option to scan sections they felt were most relevant or read all information provided. She felt it was appropriate to have separate information sheets for children and adults. The mother felt that the information provided in the child information sheet was adequate, age-appropriate and presented in an engaging format (use of colours and pictures). It was suggested that the term “psychologist” may confuse a child, so this was updated to “psychologist (talking doctor)”. It was suggested that the contact details and process for participating was made clearer on the invitation letter and parent information sheets, which were subsequently updated. The mother was also consulted with regards to which video would be best to use for children aged 9 to 13 during the tic observation measure. She suggested the Simpsons, after consulting with her son.

Appendix 37: Reasons for exclusions (recruitment stage)

Reasons participant was excluded	Retrospective referrals	New referrals
Age < 9 years old	None	7
Age > 13 years old	None	6
TS or tics were not the primary problem	4	6
FSIQ < 80	2	5
Distance (attending weekly groups impractical)	2	None
Duration or recency of previous therapy (HRT or psycho-education groups)	6	None
Tic severity too low	None	2

Note. FSIQ = Full-Scale IQ.

Appendix 38: Reasons families declined participation in the study

Reasons families declined to participate	Retrospective referrals	New referrals
Distance to travel to the groups	23	10
Treatment considered unnecessary	13	2
Child unwilling to participate in the study	12	2
Concerns regarding missing school	8	4
Financial concerns (i.e. travel costs)	7	3
Time commitment to weekly groups not possible	7	1
Parents concerned about group treatment / concerns about child developing new tics	4	1
Child not given diagnosis or family did not agree with diagnosis	4	0
Child care difficulties (e.g. for siblings)	4	0
Parents unable to take time off work	4	0
Reason not specified	3	1
Timing of groups (4.30pm) was inconvenient	2	0
Parents did not want child to be randomised	1	0
TS or tics were not considered to be the primary difficulty	1	0
Did not want to miss extracurricular activities	1	0

Note. Some families gave more than one reason for declining participation, each of which were counted independently.

Appendix 39: Observed power for significance tests (hypotheses 1 and 2)

YGTSS motor

Observed power for main effect of group type = 0.06
Observed power for main effect of time = 0.95
Observed power for interaction = 0.72

YGTSS phonic

Observed power for main effect of group type = 0.07
Observed power for main effect of time = 0.14
Observed power for interaction = 0.14

YGTSS total

Observed power for main effect of group type = 0.06
Observed power for main effect of time = 0.77
Observed power for interaction = 0.27

Tic observation

Observed power for main effect of group type = 0.08
Observed power for main effect of time = 0.80
Observed power for interaction = 0.05

Tic suppression

Observed power for main effect of group type = 0.16
Observed power for main effect of time = 0.82
Observed power for interaction = 0.09

PUTS

Observed power for main effect of group type = 0.27
Observed power for main effect of time = 0.36
Observed power for interaction = 0.05

Neuropsychological functioning: DCST

Observed power for main effect of group type = 0.15
Observed power for main effect of time = 0.05
Observed power for interaction = 0.05

Neuropsychological functioning: Flanker test

Observed power for main effect of group type = 0.06
Observed power for main effect of time = 0.18
Observed power for interaction = 0.05

Neuropsychological functioning: Fine motor dexterity

Observed power for main effect of group type = 0.10
Observed power for main effect of time = 0.06
Observed power for interaction = 0.07