The long-term effects of group-based psychological interventions for children with Tourette syndrome: A randomised controlled trial

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ABSTRACT

This randomised controlled trial examined the long-term effects of group-based psychological interventions on measures of tic severity, self-reported quality of life (QOL) and school attendance. Children ($n = 28$) with Tourette syndrome (TS) were assessed 12 months after completing a course of either group-based Habit Reversal Training (HRT) or Education. Both groups demonstrated long-term improvement in tic severity and QOL, which included significant continued improvement during the follow-up (FU) period. Both groups also showed significant post-treatment improvement in school attendance. Further research is required to explore potential therapeutic mechanisms, independent or mutual, which may underlie long-term symptom improvements.

INTRODUCTION

Tourette Syndrome (TS) is a neurodevelopmental disorder characterised by the presence of both motor and phonic tics. It is often associated with psychiatric co-morbidity, social and emotional difficulties, impaired school functioning and a diminished quality of life (Robertson, 2012; Storch et al., 2007).

Approximately 50% of individuals with TS present with a diagnosis of co-morbid obsessive compulsive disorder (OCD) or attention deficit hyperactivity disorder (ADHD; Abramovitch, Dar, Mittelman & Wilhelm, 2015; Bloch et al., 2006; Gaze, Kepley & Walkup, 2006). These co-occurring conditions can often compound the degree of functional, social and emotional impairment, impacting on the individual's overall quality of life (QOL; Specht et al., 2011; Storch et al., 2007). Indeed, research has shown that individuals with TS exhibit a significantly reduced QOL when compared to the non-TS population (Eddy et al., 2011; McGuire, Hanks, Lewin, Storch & Murphy, 2013; Müller-Vahl et al., 2010; Storch et al., 2007).

School functioning

Storch et al. (2007) noted that the presence of school functioning difficulties is a key contributor to poor self-reported QOL in children with TS. This is unsurprising given that children spend a considerable portion of their daily lives at school. Poor classroom concentration, difficulties with practical tasks (e.g. handwriting), and social isolation and bullying, have all been found to be common school-related issues experienced by children with TS (Packer, 2005; Debes, Hjalgrim & Skov, 2010). A fifth of children have been shown to experience a level of tic severity that made functioning at school at times unfeasible, affecting school attendance (Leckman et al., 1998). These findings suggest that an improvement in tic severity may lead to an improvement in school attendance rates; however, these are yet to be formally evaluated as a TS treatment outcome in children.

Treatment

Treatment guidelines endorse behavioural therapy (BT) and psychoeducation as first line interventions for tic reduction in mild to moderate TS (Steeves et al., 2012; Verdellen, van de Griendt, Hartmann, Murphy & Group, 2011). Habit Reversal Training (HRT; Azrin and Nunn, 1973) is arguably the most empirically supported behavioural approach, demonstrating medium to large treatment effects that are equivalent to effect sizes seen in drug trials of antipsychotic medication (Dutta & Cavanna, 2013; McGuire et al., 2014). Habit reversal training includes self-monitoring and awareness-building components that aim to attune the patient's awareness to the premonitory urge in order to facilitate early tic detection. Patients are then taught to apply a specific physically incompatible movement or sound termed the 'competing response', in order to effectively block the production of the tic. Habit reversal training can be combined with relaxation training and functional analysis to create a multi-component intervention termed the 'Comprehensive Behavioural Intervention for Tics' (CBIT; Woods et al., 2008).

Psychoeducational interventions aim to target impairing psychosocial and co-morbid difficulties by resolving misunderstanding around the diagnosis and alleviating anxiety (Cutler et al., 2009). Psychoeducation in a group format has been reported, in which topics included self-esteem, school, anger, attention and OCD (Murphy & Heyman, 2007). Group delivery offers the added benefit of peer support and sharing of information amongst individuals.

Building on this, adaptations have been made to behavioural treatments to broaden the focus on the individual's quality of life. Storch et al. (2012) developed a modular treatment protocol (Living with Tics) that incorporates habit reversal training with psychoeducation, problem-solving, distress tolerance and modules about coping at school, with the aim of improving tic-related impairment and resilience. Preliminary findings have highlighted the effectiveness of the Living with Tics intervention for improving QOL in children (McGuire et al., 2015).

Treatment outcomes

Evidence supports the efficacy of CBIT in reducing tic severity when delivered face-to-face as an individual treatment with both children and adults (Piacentini et al., 2010; Wilhelm et al., 2012), as well as via telehealth (Himle et al., 2012; Ricketts et al., 2016). There is also emerging evidence for the efficacy of group-based HRT, which aims to increase the availability of behavioural interventions (Yates et al., 2016).

Despite the strong body of evidence supporting behavioural therapy for TS, few studies have investigated the long-term durability of therapeutic gains following these interventions. Of those studies that have carried out follow-up (FU) assessments, Wilhelm et al. (2003) report the longest FU period (10 months) and describe a maintenance of post-treatment improvement, whilst Woods et al. (2011) found continued improvements for TS-related psychosocial symptoms at 6 months post-treatment. At present, no FUs have been conducted beyond 10 months. Due to the waxing and waning nature of tics, short-term follow-ups may indirectly capture fluctuations in the natural course of symptom presentation and longer observation periods have been recommended (Roessner et al., 2011). It should also be noted that much of the available FU data suffer high attrition rates (~30%) and have been limited to 'treatment-responders' only. A treatment-responders only methodology may not capture change in symptom severity for those who may require more time to exhibit therapeutic improvement from acute treatment. Tic severity and psychosocial functioning have typically measured outcome, but to date little attention has been paid to the school context and the impact of having tics on the child's educational life.

A recent pilot randomised controlled trial (RCT) conducted by Yates et al. (2016) evaluated short-term treatment outcomes for group-based HRT and Education in order to address the

issue of limited treatment availability and explore the feasibility of group-based treatments. Improvements in tic severity and quality of life were reported for both treatment groups, with greater tic severity improvements for the HRT group. Presently however, little is known about the long-term efficacy of group-based TS interventions.

Aims and Objectives

The current study investigated whether acute phase improvements following group treatment (Yates et al., 2016) were sustained over time, as previously shown in BT research (Verdellen et al., 2004; Wilhelm et al., 2003), by assessing 12-month follow-up outcomes. Additionally, this study aimed to investigate the relationship between TS treatments and school attendance in children. This relationship has yet to be formally evaluated; however existing research linking tic severity to school attendance (Leckman et al., 1998) indicates the potential for improvement following treatment.

METHOD

Design

This study has a cross-sectional design, collecting data at a single time point (time 3), 12 months post-treatment. The data, however, forms part of a larger longitudinal data set and was analysed together with previously collected data from time 2 (post-treatment) as reported by Yates et al. (2016). Participants were randomly allocated to either the HRT or Educational group treatment as part of the original study. Group allocation was maintained for the follow-up.

Ethical considerations

Informed consent and assent was obtained from both parents and children prior to follow-up participation. All procedures were performed in compliance with relevant laws and institutional

guidelines as approved by the hospital's research and development department, as well as all relevant ethical standards, as approved by the Queens Square Research Ethics Committee.

Participants

A total of 33 participants took part in the original trial. Participants were children that had been referred to and seen by the tic disorder clinic during the five years preceding the study. Eligibility criteria included:

- Aged 9-13 years (to coincide with the peak tic severity period);

- Primary diagnosis of TS or Chronic Tic Disorder (CTD);

- Baseline score of >13 on the Yale Global Tic Severity Scale (YGTSS; Leckman et al., 1989);

- Full-scale IQ >80.

Exclusion criteria included:

- Current or lifetime diagnosis of psychosis or substance abuse;

- Children who had previously received more than four sessions of behavioural treatment for tics;

- Children who had attended an educational session at the tic clinic within the past 2 years;

- Families whose level of English language proficiency would render it difficult to follow sessions.

Children who were successfully recruited were sequentially randomised to either HRT or Educational group treatment using an equal allocation ratio and minimization to balance age and gender. Details of the randomisation procedure and session content have previously been reported by Yates et al. (2016). The treatment interventions commenced in October 2013 and January 2014.

All children who participated in the original trial were eligible for inclusion in the follow-up study unless they had withdrawn during the trial[1]. Twenty-nine of the original study participants were contacted for follow-up recruitment and 28 participants agreed to participate[2]. Recruitment for the follow-up study took place between October 2014 and January 2015.

Outcome measures

All outcome measures were collected at all three time points, with the exception of school attendance data which was collected at pre-treatment and 12-month FU only.

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

The YGTSS is a semi-structured, clinician-administered interview involving both the child and their parent(s). It has good internal consistency, inter-rater reliability and validity in paediatric TS populations. It is considered the gold-standard measure of tic severity (Storch et al., 2005; Abramovich et al., 2015) and is the primary outcome measure for this study. Motor and phonic tics are each scored from 0 to 5 for ratings of number, frequency, intensity, complexity and interference caused in the last two weeks. Each of the two subscales can add up to a maximum of 25 points. A composite score of total tic severity (the tic severity subscale) is calculated by adding all ratings for both phonic and motor tics (with a maximum of 50 points). Children and parents are then asked about the overall impact the tics have had on the child, with scoring anchors of 0 (none), 10 (minimal), 20 (mild), 30 (moderate), 40 (marked) or 50 (severe) used to

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[1] Four participants withdrew from the original trial prior to post-treatment assessment (time 2).
[2] One participant was uncontactable having moved address and changed telephone number without informing the clinic.

indicate a score of 'impairment'. A 'total tic score' (with a maximum score of 100) can then be calculated by adding the tic severity subscale score to the impairment rating.

The Gilles de la Tourette syndrome-quality of life scale for children and adolescents (C&A-GTS-QOL; Cavanna et al., 2013).

The C&A-GTS-QOL is a 27-item measure of health-related quality of life in children with TS. It consists of four subscales (psychological, physical, obsessive-compulsive and cognitive). Items in each subscale are summed and total scores are then normalized to a 0-100 range, with higher scores indicating poorer QOL. There is also a separate 'life satisfaction' subscale (presented as a visual analogue scale) that is scored within a 0-100 range, where a higher score indicates greater life satisfaction. This study used an English translation of the measure that has been shown to have good acceptability, reliability (Cronbach's alpha > 0.7) and validity (Su et al., 2016).

School attendance

Each participant's school was contacted in order to obtain school attendance data (% attendance) for the full academic year prior to group attendance and the full academic year following group participation.

Significant life events, medication changes and further treatment

Participants were asked to provide information about any medication changes, significant life events or further psychological treatment of tics that may have occurred in the 12 months between the end of the intervention and the follow-up assessment.

Procedure

The complete battery of tests was administered by the same assessor in a single session based in a quiet room at the child's home. Assessments lasted approximately 3 hours.

All follow-up assessments were carried out approximately 12 months after the post-treatment assessment (time 2) and within 30 days of the 12-month mark, with the exception of one

participant who attended the Education group[3] (which was 14 months post Time 1). Assessments were completed by March 2015 and school attendance data collection was completed by December 2015.

Blinding

The principal assessor was not involved in any aspect of the original study and remained blind to group allocation throughout the follow-up study. Bang's Blinding Index (BI) was calculated for each treatment arm to measure success of blinding. A BI of 0.38 was calculated for the HRT condition, indicating that 38% of allocation guesses were correctly guessed beyond chance. For the Educational condition, Bang's BI was calculated as -0.15, indicating that 15% of guesses were incorrect beyond chance. Bang's blinding indices for the two treatment conditions indicate that for the majority of participants, blinding was successful, however blinding may have been unsuccessful for a few cases in the HRT condition.

Statistical analysis

A completers-only analysis ($n = 28$) was conducted in which only participants for whom data was available for both time 2 and time 3 were included. A series of 2 X 2 mixed model Repeated Measures Analysis of Variance (RM-ANOVA) tests were conducted to analyse the effects of time (T2 and T3) and group condition (HRT and Education) on tic severity and QOL, as well as any group-time interactions. School attendance data was analysed using 2 X 2 RM-ANOVA tests which examined the effects of time (T1 and T3) and group condition (HRT and Education).

Consideration of potential confounds

Tic medication changes

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[3] For this case, the time 2 assessment was delayed by 14 weeks. The time 3 assessment was conducted in line with the original time 1 assessment, and therefore 10 months after time 2.

Four participants (3 HRT, 1 Education) reported tic medication changes over the previous 12 months. Three participants had stopped taking medication due to an improvement in tic symptoms and one participant reported reducing their dosage. As tic medication has been found to significantly moderate response to CBIT and psychoeducation and supportive therapy (PST) for tics (Sukhodolsky et al., 2017), all analyses were re-run to exclude for participants that had reported a change to their medication status.

Significant life events

Seven participants reported significant life events during the follow-up period. Four participants reported family-related issues, one participant reported stress caused by school exams, one reported a depressive episode, and one participant reported being off school for three months due to physical illness. The analysis was re-run to exclude participants that reported significant life events.

Further psychological treatment

Thirteen participants engaged in psychological treatment unrelated to the study during the follow-up period. The focus of treatment varied amongst participants[4] however no participants reported engaging in further psychological therapy for tics, specifically. All analyses were re-run excluding the thirteen participants that had received further psychological treatment.

Exposure and response prevention (ERP), a therapy typically used to manage OCD symptoms, is also an evidence-based treatment for tics (Verdellen et al., 2011). Analyses were therefore also re-run to exclude the four participants (Education = 4, HRT = 0) that had received ERP for OCD. Where changes to findings were observed, these are reported.

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[5] Treatments included 3 generalised anxiety, 1 phobia, 4 OCD, 1 low mood, 2 anger, 1 'physical symptoms not related to TS' and 1 ASD

RESULTS

Twenty-eight participants took part in the follow-up study out of a total of 33 participants recruited at time 1. The five dropouts[5] did not significantly differ from completers on measures of baseline tic severity ($d = 0.18$, $p = .73$). Participants ranged in age from 10 - 14 years (mean = 12.06, $SD = 1.38$) at FU.

Baseline characteristics

Baseline characteristics are presented in Tables 1 and 2, respectively. Participants in the two conditions did not differ significantly on demographic or clinical characteristics.

Table 1 about here

Table 2 about here

Main analysis

All analyses incorporated two time points (T2 and T3) unless otherwise stated.

Tic severity

A series of four RM-ANOVA tests were conducted to test for ongoing changes in tic severity outcome variables across the FU period. These analyses used a 2 X 2 mixed model design to observe the effects of time (T2 and T3) and group condition (HRT and Education) as well as any group-time interactions. The outcome variables that were analysed included subscales of the YGTSS (motor tic severity, phonic tic severity, total tic severity) as well as the YGTSS total score. Where the assumption of sphericity has not been met, Greenhouse-Geisser corrections are reported.

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[4] The five dropouts consisted of four participants that did not complete the original study and one participant that completed the original study but was not recruited for the follow-up.

A significant effect of time was observed for motor tic severity scores ($F(1,26) = 7.52$, $p = .011$, $\eta_p^2 = .22$, $d = 1.06$) and total tic severity scores ($F(1,26) = 7.25$, $p = .012$, $\eta_p^2 = .22$, $d = 1.06$), as depicted in Figures 1 and 2.

Figure 1 about here

Figure 2 about here

No significant effects of group condition or group-time interactions were observed for either outcome measure. Complete results from the four RM-ANOVA tests are presented in Table 3.

Table 3 about here

A 2 x 2 mixed model RM-ANOVA was used to compare YGTSS tic severity outcomes for participants that attended September groups with participants that attended groups in November. Results found no significant differences between groups, indicating that the time of year that children engaged in tic treatment did not impact on tic severity outcomes.

An average 8 point reduction on the YGTSS tic severity score was observed between T1 and T3 for the HRT condition, compared to a 6 point reduction in the Educational group. These findings suggest that both groups experienced significant symptom improvement in the period between pre-treatment and follow-up, as defined by Storch et al. (2011) who recommend a 6 point change on the YGTSS tic severity score (the combination of the motor and phonic tic severity subscales) as the best indicator of treatment response.

Jeon et al (2013) propose that a 25% reduction in an individual's tic severity score represents a clinically meaningful change or a 'responder'. In accordance with this proposed benchmark, 46.2% ($n = 6$; range of tic reduction = -12.5% to 63.0%) of participants in the Educational group would be considered treatment "responders", whilst 53.3% ($n = 8$; range of tic reduction = -

23.8% to 70.6%) would be considered "responders" in the HRT group, when measuring symptom change between T1 and T3. A smaller proportion of participants were classified as treatment 'responders' at T2 (18% of Educational group participants and 35% of HRT participants), indicating continued improvement throughout the follow-up period.

Quality of life

GTS-QOL total scores were analysed using a 2 x 2 RM-ANOVA test. A mixed model design was implemented with group condition (HRT and Education) as the between-subjects factor and time (T2 and T3) as the within-subjects factor. There were no main effects of time ($F(1,26) = 0.01$, $p = .942$), group condition ($F(1,26) = 1.577$, $p = .220$) or group-time interaction ($F(1,26) = 1.05$, $p = .315$).

A secondary analysis was conducted excluding cases that had reported significant life events during the follow-up period ($n = 7$), in order to account for the potential negative impact of external life events unrelated to diagnosis, on QOL outcomes. Results of this analysis showed a significant main effect of time ($F(1,19) = 5.73$, $p = .027$, $\eta_p^2 = .23$, $d = 1.09$). These findings indicate that both groups demonstrated continued improvement in QOL during the 12 months after completing group treatment, with a 7 point mean improvement observed in the Educational condition whilst the HRT group improved by a mean of 3 points. Results from the RM-ANOVA test are depicted in Figure 3.

Figure 3 about here

A 2 x 2 mixed model RM-ANOVA was used to compare participants that attended September groups with participants attending November groups (excluding those with significant recent life events, as above). Results found no significant differences between groups, indicating that the time of year that children engaged in treatment did not impact on improvement in QOL total scores.

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School attendance

The HRT group reported a change in attendance rates from 92.34% (pre-treatment) to 95.22% (post-treatment), whilst there was also an observed shift from 93.69% to 95.99% in the Educational group.

A main effect of time was observed ($F(1,23) = 10.04$, $p = .004$, $\eta_p^2 = .30$, $d = 1.31$). There was no main effect of group ($F(1,23) = 0.13$, $p = .723$) or group-time interaction ($F(1,23) = 0.51$, $p = .484$). These results indicate that children in both group conditions demonstrated significantly higher school attendance rates at post-treatment when compared to pre-treatment school attendance (see Figure 4).

Figure 4 about here

DISCUSSION

Main findings

This study investigated 12-month follow-up outcomes of group interventions for children with TS. Overall, findings offer support for the long-term efficacy of group-based HRT and Educational treatments. Results highlight continued improvements in tic severity in both HRT and Educational group participants over the follow-up period, with indication of a slightly greater improvement in HRT participants. Both groups demonstrated continued improvement in QOL over the follow-up period. Results also indicated that children in both group conditions improved in school attendance.

Tic severity

Twelve months post-treatment, children in both group conditions maintained the tic severity improvements observed following the acute phase. In addition, both groups demonstrated continued tic severity improvements. These appear to be predominantly driven by ongoing

improvements in motor tic severity, as observed on the motor subscale of the YGTSS. There is a tentative suggestion of greater motor tic improvement amongst the HRT group compared to the Educational group, with the HRT condition demonstrating a 17.2% improvement in motor tic severity across the follow-up period, compared to an 11.4% improvement in the Educational group. This potential group difference in improvement across the follow-up period is in line with group differences observed immediately following the acute phase (Yates et al., 2016).

Findings for the HRT group in this study are comparable to previous trials, although on a more modest scale. In a trial comparing CBIT to supportive psychotherapy and education, Piacentini et al. (2010) report a 11.4-point tic severity reduction between pre-treatment and six-month follow-up in the CBIT condition. The present study found a smaller reduction of 7.7 points for the behavioural intervention. It is possible that this study's more modest findings reflect a diluting effect of group-based behavioural treatments compared to individual treatment. It should however be noted that Piacentini et al.'s study describes a shorter FU period when compared to the present study. Furthermore, Piacentini et al.'s 6-month follow-up analysis consisted of 'treatment-responders' only. This could explain the comparatively smaller reduction in tic severity observed in the present FU study, which included all participants with a complete data set. The educational group findings from this study demonstrated a 5.9-point reduction in tic severity, which would be considered an indication of long-term response to treatment, in line with Storch et al. (2011)'s guideline of a 6-point reduction as optimal indication of treatment response.

It should be noted that other follow-up studies conducted by Verdellen et al. (2004) and McGuire et al. (2015) have offered longer interventions, reporting treatment lengths of 12 and 10 sessions, respectively. Future studies aiming to replicate the findings of this study would benefit from increasing the number of treatment sessions. This would enable more confident comparisons to be made between group and individual interventions.

Continued improvement in tic severity was not anticipated or hypothesised. Of the follow-up data that exists, findings have predominantly shown a stabilisation of treatment effects, maintained between post-treatment and follow-up assessments (Piacentini et al., 2010; Wilhelm et al., 2003). There are several theoretical explanations for this continued long-term improvement. Firstly, it should be noted that thirteen participants engaged in further psychological treatment during the follow-up period (7 HRT; 6 Education). Although in all cases on going intervention was not directly focused on tic symptoms, it is possible that an improvement in related conditions, such as OCD or generalised anxiety, may positively influence tic symptoms. Indeed, anxiety has been found to exacerbate tic severity (Conelea & Woods, 2008), which suggests that alleviating anxiety may consequently contribute to tic severity improvement in the long-term.

Another theory is the possibility of two distinct therapeutic working mechanisms responsible for tic improvement in each of the two groups. Children randomised to the HRT group may have continued to practice and master tic suppression strategies following group completion, leading to ongoing improvements.

Continued tic severity improvements observed for the Educational group could be hypothesised as a secondary outcome of the long-term beneficial effects of psychoeducation on managing psychosocial symptoms such as anxiety and school functioning (Nussey, Pistrang and Murphy, 2014).

It could also be theorised that a mutual underlying mechanism was present in both groups, hence influencing tic severity symptoms in both conditions. Examples of this include exposure to peer support, shared coping strategies and social normalisation of symptoms. Again, this could lead to a reduction in overall anxiety that could positively influence tic severity and QOL.

It should be noted that there were elements of overlap between the two group intervention protocols. Both groups included psychoeducation about Tourette syndrome, a relaxation

technique and use of reward strategies. It is possible that these shared therapeutic components may have contributed to the similar rates of improvement observed between the two groups.

Quality of life

Participants in HRT and Educational groups maintained QOL improvements at FU after excluding participants that had experienced a recent significant life event. Furthermore, continued long-term improvements were observed for both groups of participants across the FU period, with no significant differences between conditions. This continued improvement is consistent with previous research suggesting that psychosocial outcomes show greater improvement at six-month follow-up than directly following individual tic treatment, indicating that participants may benefit from a consolidation period following intervention (Woods et al., 2011). Similar long-term continued QOL improvement has been demonstrated in studies evaluating cognitive behavioural therapy (CBT) for social anxiety (Watanabe et al. 2010), as well as in long-term anxiety outcomes following brief CBT (Crawley et al., 2013).

Findings demonstrated no differences in long-term QOL improvement between September and November groups. This finding is contrary to group differences observed at T2 (Yates et al., 2016) and suggests attenuation of any short-term disadvantages to beginning treatment earlier in the academic year.

School attendance

Participants in both conditions experienced a rise in school attendance to just above 95% following group treatment. Importantly, these findings place both groups of participants in line with the national secondary school average of 95% (Department of Education, 2016a). These findings are particularly significant in the context of national statistics that show a 0.8% increase in authorised absences between the ages of 10 and 12 years (Department of Education, 2016b). It could be theorised that the observed improvement in school attendance may be associated

with the demonstrated significant improvements in tic severity between T1 and T3. As previously reported, Leckman et al. (1998) identified that up to 20% of children with TS may experience school-interfering tic severity, affecting their ability to attend school. This suggests that an improvement in tic severity may directly reduce some of the tic-related difficulties experienced in the classroom, which could subsequently improve school attendance.

The group format of the interventions may have, to some extent, simulated a classroom setting, with children 'taught' in the presence of other children. This format could therefore offer specific therapeutic gains that are more readily transferable to a school setting. Future studies could directly compare school attendance outcomes of individual and group treatments in order to clarify whether there is a unique benefit of group-based interventions.

Clinical implications

These findings suggest that group treatment for TS can be considered a reasonable alternative to individual treatment where services are resource-limited. This study also offers specific support for the provision of group-based education as an alternative to behavioural treatment, with comparable long-term outcomes. These findings contribute to a wider evidence base that can support clinicians and families to make informed decisions about treatment. Results indicate that group treatments can contribute not only to reductions in tic severity, but can also positively impact on a child's school attendance and self-perceived quality of life in the long-term.

Strengths and limitations

A detailed description of the strengths and limitations of the original study design has been reported previously (Yates et al., 2016). In brief, the strengths of this study lay predominantly in the single-blinded, randomised, controlled design. This design allowed for the robust measurement of clinical change over time in two group conditions that were matched for frequency, duration and clinician-contact time. Treatment in both groups was structured and protocol-driven to maintain treatment fidelity and outcome measures were scored using the same

detailed scoring protocols across all three time points to maintain consistency. Treatment was conducted by highly experienced clinicians within a specialist clinic setting. A gold standard clinician-rated tic severity scale (YGTSS) was used as the main outcome measure, and evaluator blindness was measured and deemed acceptable.

The study had an overall attrition rate of 15% between T1 and T3. The follow-up study benefitted from low attrition (3.4%), with only one participant lost between T2 and T3. This is significantly lower than in previously reported follow-ups (Verdellen et al., 2004; Wilhelm et al., 2012), and may have benefitted from the use of home-based assessments, increasing convenience for the family and reducing the burden of travelling to the clinic. Home-based assessments have been found to highly correlate with clinic observations (Himle et al., 2006; Piacentini et al., 2006) and allow for the measurement of symptoms in an environment familiar to the child, increasing the ecological validity of the measures.

The lack of an additional control group limits the conclusions that can be made from the findings. Firstly, although the rater was blind to the treatment allocation, neither the rater nor the participants were blind to the fact that all participants had received some form of treatment. This may have contributed to rater-bias for both the self-report measures and the YGTSS clinician-rated measure. Secondly, as previously discussed, natural remission of symptoms may have occurred over the course of the 12-month follow-up period. A waitlist control group could allow for this natural effect to be monitored and controlled for, enabling clarification of any additional treatment effects. Mean age at T1 and T3 assessments did however fall within or very close to the peak severity period of between 10-12 years (Bloch et al., 2006; Leckman et al., 1998), suggesting that significant, naturally occurring tic severity reductions are unlikely to have occurred amongst the sample. This study used opportunity sampling as a recruitment method and it is possible that patients experiencing peak tic severity would be particularly inclined to participate in the study. This may have made the sample of participants particularly susceptible to post-study natural symptom remission. Either a waitlist control group or the use of systematic

consecutive sampling taken from the clinic's referral flow in place of opportunity sampling may have reduced this effect.

Future research

Given the small sample size of the current study, a larger RCT would be recommended in order to replicate these findings and explore further predictors of long-term treatment response that this study was not sufficiently powered to investigate. A multi-site, community-based study would also be informative. Future studies would benefit from a waitlist control in order to account for natural symptom change, or a 'minimal treatment' condition to account for the potential placebo effect of perceived support from a specialist clinic. It would be interesting to include an alternative control group consisting of individually administered treatment in order to directly measure the effect of a group-based format. This would help determine whether there are any presiding long-term benefits of group treatment over individual treatment.

Given that both the HRT and Educational groups demonstrated significant symptom improvement in both the short-term and the long-term, further investigation into the precise mechanisms of change in these respective treatments (and to determine whether these therapeutic mechanisms are mutual or distinguishable between the two approaches) would be important for the future design and development of TS interventions.

Conclusion

In a sample of children with diagnosed TS and chronic tic disorders, group-based interventions offered sustained benefits to tic severity, quality of life and school attendance, with continued improvement observed in both group conditions over the follow-up period. These results support the implementation of HRT and Educational group treatments for children with TS. Future studies would benefit from a larger sample and alternative control groups.

REFERENCES

Abramovitch, A., Dar, R., Mittelman, A., & Wilhelm, S. (2015). Comorbidity between attention deficit/hyperactivity disorder and obsessive-compulsive disorder across the lifespan: a systematic and critical review. *Harvard review of psychiatry*, *23*(4), 245-262.

Azrin, N. H., & Nunn, R. G. (1973). Habit-reversal: a method of eliminating nervous habits and tics. *Behaviour research and therapy*, *11*(4), 619-628.

Bloch, M. H., Peterson, B. S., Scahill, L., Otka, J., Katsovich, L., Zhang, H., & Leckman, J. F. (2006). Adulthood outcome of tic and obsessive-compulsive symptom severity in children with Tourette syndrome. *Arch Pediatr Adolesc Med, 160*(1), 65-69.

Cavanna, A. E., Luoni, C., Selvini, C., Blangiardo, R., Eddy, C. M., Silvestri, P. R., … Cardona, F. (2013). The Gilles de la Tourette Syndrome-Quality of Life Scale for children and adolescents (C&A-GTS-QOL): Development and validation of the Italian version. *Behavioural Neurology*, *27*(1), 95–103.

Conelea, C. A., & Woods, D. W. (2008). The influence of contextual factors on tic expression in Tourette's syndrome: A review. *Journal of Psychosomatic Research, 65*(5), 487-496.

Crawley, S. A., Kendall, P. C., Benjamin, C. L., Brodman, D. M., Wei, C., Beidas, R. S., ... & Mauro, C. (2013). Brief cognitive-behavioral therapy for anxious youth: Feasibility and initial outcomes. *Cognitive and behavioral practice*, *20*(2), 123-133.

Cutler, D., Murphy, T., Gilmour, J., & Heyman, I. (2009). The quality of life of young people with Tourette syndrome. *Child: Care, health and development*, *35*(4), 496-504.

Debes, N., Hjalgrim, H., & Skov, L. (2010). The presence of attention-deficit hyperactivity disorder (ADHD) and obsessive-compulsive disorder worsen psychosocial and educational problems in Tourette syndrome. *Journal of child neurology*, *25*(2), 171-181.

Deckersbach, T., Rauch, S., Buhlmann, U., & Wilhelm, S. (2006). Habit reversal versus supportive psychotherapy in Tourette's disorder: a randomized controlled trial and

predictors of treatment response. *Behav Res Ther, 44*(8), 1079-1090.

Department of Education. (2016a). *Pupil absence in schools in England: Autumn Term 2015* (Report

No. SFR13). Retrieved from

https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/5240

81/SFR13_2016_Text.pdf

Department of Education. (2016b). *Pupil absence in schools in England: 2014 to 2015* (Report No.

SFR10). Retrieved from

https://www.gov.uk/government/statistics/pupil-absence-in-schools-in-england-2014-

to-2015

Dutta, N., & Cavanna, A. E. (2013). The effectiveness of habit reversal therapy in the treatment

of Tourette syndrome and other chronic tic disorders: a systematic review. *Functional

neurology, 28*(1), 7.

Eddy, C. M., Cavanna, A. E., Gulisano, M., Agodi, A., Barchitta, M., Cali, P., . . . Rizzo, R.

(2011). Clinical correlates of quality of life in Tourette syndrome. *Mov Disord, 26*(4), 735-

738.

Gaze, C., Kepley, H. O., & Walkup, J. T. (2006). Co-occurring psychiatric disorders in children

and adolescents with Tourette syndrome. *J Child Neurol, 21*(8), 657-664.

Himle, M. B., Chang, S., Woods, D. W., Pearlman, A., Buzzella, B., Bunaciu, L., & Piacentini, J.

C. (2006). Establishing the feasibility of direct observation in the assessment of tics in

children with chronic tic disorders. *Journal of applied behavior analysis, 39*(4), 429-440.

Himle, M. B., Freitag, M., Walther, M., Franklin, S. A., Ely, L., & Woods, D. W. (2012). A

randomized pilot trial comparing videoconference versus face-to-face delivery of

behavior therapy for childhood tic disorders. *Behav Res Ther, 50*(9), 565-570.

Jeon, S., Walkup, J. T., Woods, D. W., Peterson, A., Piacentini, J., Wilhelm, S., ... & Scahill, L.

(2013). Detecting a clinically meaningful change in tic severity in Tourette syndrome: a

comparison of three methods. *Contemporary clinical trials, 36*(2), 414-420.

Leckman, J. F., Riddle, M. A., Hardin, M. T., Ort, S. I., Swartz, K. L., Stevenson, J. O. H. N., & Cohen, D. J. (1989). The Yale Global Tic Severity Scale: initial testing of a clinician-rated scale of tic severity. *Journal of the American Academy of Child & Adolescent Psychiatry*, *28*(4), 566-573.

Leckman, J. F., Zhang, H., Vitale, A., Lahnin, F., Lynch, K., Bondi, C., . . . Peterson, B. S. (1998). Course of tic severity in Tourette syndrome: the first two decades. *Pediatrics, 102*(1 Pt 1), 14-19.

McGuire, J. F., Arnold, E., Park, J. M., Nadeau, J. M., Lewin, A. B., Murphy, T. K., & Storch, E. A. (2015). Living with tics: reduced impairment and improved quality of life for youth with chronic tic disorders. *Psychiatry Res, 225*(3), 571-579.

McGuire, J. F., Hanks, C., Lewin, A. B., Storch, E. A., & Murphy, T. K. (2013). Social deficits in children with chronic tic disorders: phenomenology, clinical correlates and quality of life. *Compr Psychiatry, 54*(7), 1023-1031.

McGuire, J. F., Piacentini, J., Brennan, E. A., Lewin, A. B., Murphy, T. K., Small, B. J., & Storch, E. A. (2014). A meta-analysis of behavior therapy for Tourette syndrome. *Journal of Psychiatric Research*, *50*, 106-112.

Muller-Vahl, K., Dodel, I., Muller, N., Munchau, A., Reese, J. P., Balzer-Geldsetzer, M., . . . Oertel, W. H. (2010). Health-related quality of life in patients with Gilles de la Tourette's syndrome. *Mov Disord, 25*(3), 309-314.

Murphy, T., & Heyman, I. (2007). Group work in young people with Tourette Syndrome. *Child and Adolescent Mental Health*, *12*(1), 46-48.

Nussey, C., Pistrang, N., & Murphy, T. (2014). Does it help to talk about tics? An evaluation of a classroom presentation about Tourette syndrome. *Child and Adolescent Mental Health*, *19*(1), 31-38.

Packer, L. E. (2005). Tic-related school problems: impact on functioning, accommodations, and interventions. *Behavior Modification*, *29*(6), 876-899.

Piacentini, J., Himle, M. B., Chang, S., Baruch, D. E., Buzzella, B. A., Pearlman, A., & Woods, D. W. (2006). Reactivity of tic observation procedures to situation and setting. *Journal of Abnormal Child Psychology*, *34*(5), 647-656.

Piacentini, J., Woods, D. W., Scahill, L., Wilhelm, S., Peterson, A. L., Chang, S., . . . Walkup, J. T. (2010). Behavior therapy for children with Tourette disorder: a randomized controlled trial. *JAMA, 303*(19), 1929-1937

Ricketts, E. J., Goetz, A. R., Capriotti, M. R., Bauer, C. C., Brei, N. G., Himle, M. B., . . . Woods, D. W. (2016). A randomized waitlist-controlled pilot trial of voice over Internet protocol-delivered behavior therapy for youth with chronic tic disorders. *J Telemed Telecare, 22*(3), 153-162.

Robertson, M. M. (2012). The Gilles de la Tourette syndrome: the current status. *Archives of disease in childhood-Education & practice edition*, *97*(5), 166-175.

Roessner, V., Plessen, K. J., Rothenberger, A., Ludolph, A. G., Rizzo, R., Skov, L., ... & ESSTS Guidelines Group. (2011). European clinical guidelines for Tourette syndrome and other tic disorders. Part II: pharmacological treatment. *European child & adolescent psychiatry*, *20*(4), 173-196.

Specht, M. W., Woods, D. W., Piacentini, J., Scahill, L., Wilhelm, S., Peterson, A. L., . . . Walkup, J. T. (2011). Clinical Characteristics of Children and Adolescents with a Primary Tic Disorder. *J Dev Phys Disabil, 23*(1), 15-31.

Steeves, T., McKinlay, B. D., Gorman, D., Billinghurst, L., Day, L., Carroll, A., . . . Pringsheim, T. (2012). Canadian guidelines for the evidence-based treatment of tic disorders: behavioural therapy, deep brain stimulation, and transcranial magnetic stimulation. *Can J Psychiatry, 57*(3), 144-151.

Storch, E. A., De Nadai, A. S., Lewin, A. B., McGuire, J. F., Jones, A. M., Mutch, P. J., ... & Murphy, T. K. (2011). Defining treatment response in pediatric tic disorders: a signal detection analysis of the Yale Global Tic Severity Scale. *Journal of child and adolescent*

psychopharmacology, 21(6), 621-627.

Storch, E. A., Merlo, L. J., Lack, C., Milsom, V. A., Geffken, G. R., Goodman, W. K., & Murphy, T. K. (2007). Quality of life in youth with Tourette's syndrome and chronic tic disorder. *J Clin Child Adolesc Psychol, 36*(2), 217-227.

Storch, E. A., Morgan, J. E., Caporino, N. E., Brauer, L., Lewin, A. B., Piacentini, J., & Murphy, T. K. (2012). Psychosocial treatment to improve resilience and reduce impairment in youth with tics: an intervention case series of eight youth. *Journal of Cognitive Psychotherapy, 26*(1), 57-70.

Storch, E. A., Murphy, T. K., Geffken, G. R., Sajid, M., Allen, P., Roberti, J. W., & Goodman, W. K. (2005). Reliability and validity of the Yale Global Tic Severity Scale. *Psychol Assess, 17*(4), 486-491.

Su, M. T., McFarlane, F., Cavanna, A. E., Termine, C., Murray, I., Heidemeyer, L., ... & Murphy, T. (2017). The English Version of the Gilles de la Tourette Syndrome–Quality of Life Scale for Children and Adolescents (C&A-GTS-QOL) A Validation Study in the United Kingdom. *Journal of child neurology, 32*(1), 76-83.

Sukhodolsky, D. G., Woods, D. W., Piacentini, J., Wilhelm, S., Peterson, A. L., Katsovich, L., ... & Scahill, L. (2017). Moderators and predictors of response to behavior therapy for tics in Tourette syndrome. *Neurology, 88*(11), 1029-1036.

Verdellen, C., van de Griendt, J., Hartmann, A., Murphy, T., & Group, E. G. (2011). European clinical guidelines for Tourette syndrome and other tic disorders. Part III: behavioural and psychosocial interventions. *Eur Child Adolesc Psychiatry, 20*(4), 197-207.

Verdellen, C. W., Keijsers, G. P., Cath, D. C., & Hoogduin, C. A. (2004). Exposure with response prevention versus habit reversal in Tourettes's syndrome: a controlled study. *Behav Res Ther, 42*(5), 501-511.

Watanabe, N., Furukawa, T. A., Chen, J., Kinoshita, Y., Nakano, Y., Ogawa, S., ... & Noda, Y. (2010). Change in quality of life and their predictors in the long-term follow-up after

group cognitive behavioral therapy for social anxiety disorder: a prospective cohort study. *BMC psychiatry*, *10*(1), 81.

Wilhelm, S., Deckersbach, T., Coffey, B. J., Bohne, A., Peterson, A. L., & Baer, L. (2003). Habit reversal versus supportive psychotherapy for Tourette's disorder: a randomized controlled trial. *Am J Psychiatry, 160*(6), 1175-1177.

Wilhelm, S., Peterson, A. L., Piacentini, J., Woods, D. W., Deckersbach, T., Sukhodolsky, D. G., ... & Scahill, L. (2012). Randomized trial of behavior therapy for adults with Tourette syndrome. *Archives of general psychiatry*,*69*(8), 795-803.

Woods, D. W. (2008). *Managing Tourette syndrome: A behavioral intervention for children and adults therapist guide*. USA: Oxford University Press.

Woods, D. W., Piacentini, J. C., Scahill, L., Peterson, A. L., Wilhelm, S., Chang, S., . . . Walkup, J. T. (2011). Behavior therapy for tics in children: acute and long-term effects on psychiatric and psychosocial functioning. *J Child Neurol, 26*(7), 858-865.

Yates, R., Edwards, K., King, J., Luzon, O., Evangeli, M., Stark, D., ... & Murphy, T. (2016). Habit reversal training and educational group treatments for children with tourette syndrome: A preliminary randomised controlled trial. *Behaviour research and therapy*, *80*, 43-50.