

Habit Reversal Training and Educational group treatments for children with Tourette syndrome: a preliminary randomised controlled trial

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Abstract

Quality of life of children with Tourette Syndrome (TS) is impacted greatly by its symptoms and their social consequences. Habit Reversal Training (HRT) is effective but has not, until now, been empirically evaluated in groups.

This randomised controlled trial evaluated feasibility and preliminary efficacy of eight HRT group sessions compared to eight Education group sessions. Thirty-three children aged 9 to 13 years with TS or Chronic Tic Disorder took part. Outcomes evaluated were tic severity and quality of life (QoL).

Tic severity improvements were found in both groups. Motor tic severity (Yale Global Tic Severity Scale) showed greatest improvements in the HRT group. Both groups showed a strong tendency toward improvements in patient reported QoL.

In conclusion, group-based treatments for TS are feasible and exposure to other children with tics did not increase tic expression. HRT led to greater reductions in tic severity than Education. Implications, such as cost-effectiveness of treatment delivery, are discussed.

Keywords: Tourette syndrome; Group; Habit Reversal Training; Education; Randomised Controlled Trial; Children

Abbreviations: CI = Confidence interval; CMTD = Chronic Motor Tic Disorder

Introduction

Tourette Syndrome (TS) is a developmental neuropsychiatric disorder defined by multiple motor tics and at least one vocal tic present for over a year (American Psychiatric Association, 2013). TS has a prevalence of 0.7% among UK 13-year-olds (Scharf, Miller, Mathews & Ben-Shlomo, 2012) and is four times more common in males (Freeman et al., 2000). Tics tend to fluctuate, occurring in bouts over time (Leckman et al., 1998) and symptoms peak between 10 and 12 years (Bloch & Leckman, 2009). The condition is associated with high comorbidity with Attention Deficit Hyperactivity Disorder (ADHD) and Obsessive Compulsive Disorder (OCD).

Children with TS report functional impairment (Storch et al., 2007) and diminished Quality of Life (QoL; Cutler, Murphy, Gilmour, & Heyman, 2009). The impact of having TS can continue into adulthood (Lewin et al., 2012).

There is good evidence supporting behavioural therapy for TS. Recent meta-analyses indicate that Habit Reversal Training (HRT) has the best empirical support for reducing tic severity (McGuire et al., 2014). HRT helps participants develop an awareness of when tics are about to occur. The individual then develops a behaviour to stop the tic when the urge to tic arises. Through practice, increased control is gained over each troublesome tic. HRT has been developed into a manualised Comprehensive Behavioural Intervention for Tics (CBIT; Woods et al., 2008). The eight-session treatment includes a relaxation component and additional functional analysis to minimise environmental triggers. CBIT has been evaluated for use with children in a large scale randomised controlled trial (RCT) and showed greater tic reduction when compared to “supportive psychotherapy and education”, with a medium effect size (Cohen's $d = 0.68$; Piacentini et al., 2010). Six months later “responders” demonstrated reductions in anxiety and disruptive behaviour and improved social functioning (Woods et al., 2011). HRT and its variants show tic reductions of 30 to 100% (Verdellen, van de Griendt, Hartmann, & Murphy, 2011) and medium to large effect sizes relative to control groups (McGuire et al., 2014). Studies examining

effects of HRT on phonic and motor tics separately have reported mixed results and there is no clear pattern to date showing which tics respond most to treatment (Piacentini et al., 2010; Wilhelm et al., 2012).

Verdellen, et al. (2011) argue that psycho-education could reduce uncertainty about the condition and self-stigma.

Group-based HRT has not yet been empirically evaluated, but could provide an additional option as a cost-effective treatment for large numbers. Group-based delivery may provide additional benefits, such as improved self-efficacy, reduced isolation and help children explain their symptoms to peers (Murphy & Heyman, 2007; Nussey, Pistrang & Murphy, 2014).

The current study investigated feasibility and preliminary efficacy of HRT and Education groups for children aged 9 to 13 years with TS or Chronic Tic Disorder (CTD). The groups were evaluated in terms of tic severity and QoL outcomes. It was predicted that the HRT group would experience greater reductions in tic severity compared to the Education group, as tics were the direct focus of the intervention. Children in both groups were predicted to show significant post-treatment QoL improvements, as each treatment addresses different factors impacting QoL.

Method

The study was a single-blind RCT, reviewed and approved by London Queen Square Research Ethics Committee and by ethics committees for Royal Holloway, University of London and University College London. The trial is registered on the National Institute for Health Research Portfolio Database (ISRCTN 50798741, <http://www.controlled-trials.com>).

Recruitment was from a specialist TS clinic at a London hospital where participants had received a diagnosis of TS or CTD from an experienced multidisciplinary team. The recruitment period was predefined (June to November 2013). All children aged 9 to 13 years, assessed within the preceding five years, were invited to participate. Additionally, children referred during

recruitment were invited if they met inclusion criteria. Each child was randomised to either an HRT or an Education group. Both groups were described to participants as active interventions with potential but unknown benefits. Pre-treatment assessments took place during the month prior to treatment (Time 1) and post-treatment assessments within a month of treatment end (Time 2). All assessments took place in participants' homes with the exception of four pre-assessments which, for practical reasons, took place in the clinic. Participants were excluded if they had: a Yale Global Tic Severity Scale (YGTSS; Leckman et al., 1989) total tic severity score < 13; a Full Scale IQ < 80; insufficient spoken English to participate in treatment; attended an Education group at the clinic within the previous two years; attended more than four individual HRT sessions or if TS was not the primary presenting problem. Figure 1 shows the progress of participants through the study.

[Insert Figure 1 here]

Having given informed consent, participants were sequentially randomised to treatment group using an equal allocation ratio. Minimisation software maximally balanced age and gender across conditions (Treasure & MacRae, 1998). Assessors were blind to treatment condition. Following data entry, Bang's blinding indices were calculated (Bang, Ni, & Davis, 2004), representing the proportion of unblinding occurring. In the HRT condition 35% of condition assignments were correctly guessed by the researchers, beyond chance. In the Education condition there was a slight tendency for the researchers to incorrectly guess that participants had been assigned to HRT (12.5%).

Interventions

One HRT and one Education group ran from September to October 2013. A second of each group type ran from November 2013 to January 2014. Participants attended only the eight-session group to which they were randomised and received no individual TS related sessions.

Participants received ongoing treatment-as-usual in terms of school liaison and medication. Alongside the children's groups, parents were invited to attend four parent sessions.

All parent and child group sessions were structured and manualised (available on request). The core therapeutic content differed between the groups, but practical elements were similar. The initial two sessions lasted 90 minutes and remaining sessions an hour. Sessions took place at the clinic and were run by five clinicians (three qualified and two in training). All clinicians were trained in delivery of the group protocol by author TM, who facilitated the children's groups.

Sessions involved group discussion, didactic teaching and small group activities. A small weekly homework task was completed with parental support. Both groups started with the same first session of education about tics and both included teaching on progressive muscle relaxation. Reward strategies were used to increase implementation of techniques learnt.

Fidelity to treatment manuals was monitored using a fidelity checklist similar to those used in previous studies (e.g. Sukhodolsky et al., 2009). The approach resulted in complete fidelity to the protocols except that several sessions ran out of time in the final few minutes, across both conditions.

HRT group. This protocol was based on individual HRT treatment for children with TS (Woods et al., 2008) and an HRT therapy manual and workbook developed by Verdellen, van de Griendt, Kriens and van Oostrum (2011). The children chose up to three tics to treat, of which 73% were motor tics. Details of the tics chosen by each participant are given in Table 1. The most bothersome tics were selected and treated first and skills developed to apply to further tics. Following specific, detailed instruction on competing responses, participants worked in small groups, with support from clinicians to develop and evaluate competing responses for each chosen tic.

[Insert Table 1 here]

Education group. This protocol was based on a six-session psycho-educational intervention (Murphy & Heyman, 2007), adapted to increase structural similarity to the HRT intervention. The content of each session was: Tics and TS; Self-esteem; School; Anger; Anxiety and OCD; Attention; Planning and Organising; Review, Quiz and Certificates. Sessions used cognitive behavioural strategies, such as identification of triggers to anger and problem solving approaches to build self-esteem and organisational skills. No instructions on managing tics were provided.

Parent groups. Sessions followed a similar structure to the children's groups including homework review and support, written handouts, group discussions, group-specific content (linked to the content of the children's groups) and implementation of reward strategies. No instructions on managing tics were provided.

Measures

The YGTSS (Leckman et al., 1989) is considered the gold standard tic severity measure. A list is generated of motor and phonic tics present over the past week, followed by ratings of number, frequency, intensity and complexity of tics and degree of interference caused. The primary outcome measures were YGTSS composite scores of motor and phonic tic severity (both rated 0 to 25). Twenty percent of the videos were randomly selected and double coded. Good inter-rater reliability was shown for both motor (ICC = .88; 95% CI: .62, .97) and phonic (ICC = .95; 95% CI: .83, .99) tic severity.

The Gilles de la Tourette Syndrome Quality of Life Scale for Children and Adolescents (GTS-QOL C&A; Cavanna et al., 2013) is the only condition-specific measure of health-related QoL for children with TS. This 27-item self-report measure reflects psychological, physical and cognitive elements of QoL. The total score of an unpublished English version of the measure was used. The measure showed high internal consistency ($\alpha = .89$).

To characterise the groups at baseline, ADHD symptoms were measured using the MTA parent-report version of Swanson, Nolan, and Pelham–IV (SNAP-IV; Swanson et al., 2001). OCD symptoms were measured using the parent-report version of the Children’s Obsessive Compulsive Inventory Revised (ChOCI-R; Uher, Heyman, Turner, & Shafran, 2008). A short form of the Wechsler Intelligence Scales for Children – IV (Crawford, Anderson, Rankin, & MacDonald, 2010) was used to estimate Full Scale IQ (FSIQ).

Analysis

All participants remained in their assigned groups. An intention-to-treat (ITT) analysis was used. All participants assessed at baseline ($n = 33$) were included, using last-observation carried forward for those lost to post-treatment assessment ($n = 4$). A subsequent secondary analysis included only participants who attended five or more sessions ($n = 26$) to provide a measure of effects when the protocol was adhered to.

Hypotheses were tested using repeated measures Analysis of Variance (ANOVA) tests. A 2x2 mixed design examined main effects of group (HRT or Education) as a between-subjects factor and time-point as a within-subjects factor, as well as interaction effects.

Results

Tables 2 and 3 display descriptive data for each group and for the full sample. They also show results of tests for baseline group differences. No significant group differences in baseline characteristics were found on any variables.

[Insert Tables 2 and 3]

Four children received ongoing therapy (Cognitive Behavioural Therapy or counselling) for other conditions (anger, anxiety or mood) during the study (HRT $n = 3$; Education $n = 1$). Five had medication changes between baseline and post-treatment (HRT $n = 3$; Education $n = 2$).

Three participants were in the HRT group and two participants in the psycho-education group. In the HRT group one child began taking Sertraline about a week after the preassessment and then stopped again three weeks before follow-up. A second child had a 50% dosage increase in epilepsy medication towards the end of the group sessions and before follow-up. The third child had a 17% reduction in their dose of stimulant medication (methylphenidate) during this time. In the psycho-education group two children stopped taking non-stimulant medication between pre-assessment and follow-up. These changes involved both increases and reductions in medication so any influence on results is likely to have been balanced. Nonetheless, the analyses were repeated excluding these children with no significant changes seen in the results.

One child in each group experienced stressful events (illness of a relative during the group and another child experienced symptoms following completion of the intervention before post-treatment assessment). The analysis was run both including and excluding these participants. Where the overall pattern of results was significantly altered these differences are discussed below.

Tic Outcomes

Repeated measures ANOVAs were separately conducted for YGTSS motor and phonic tic severity. Means and standard deviations are reported in Table 4. For motor tics, the main effect of group was non-significant ($F(1,31) = 0.054, p = .818$). The main effect of time-point was significant ($F(1,31) = 13.87, p = .001$, Cohen's $d = 1.58$, large effect), with participants in both groups reporting reductions in motor tics. The means (SDs) reduced from 16.98 (0.70) at baseline to 15.50 (0.60). A significant interaction was found between time-point and group ($F(1,31) = 6.90, p = .013$, adjusted Cohen's $d = 0.55$, medium effect), suggesting a significantly larger reduction in motor tics in the HRT group. In the Education group scores fell from 16.31

(3.03) to 15.88 (2.28), whereas in the HRT group they fell from 17.65 (4.74) to 15.12 (4.30). In contrast, for the phonic tic scale, the main effects of both group ($F(1,31) = 0.179, p = .675$) and time-point ($F(1,31) = 0.821, p = .372$) were non-significant, as was the interaction ($F(1,31) = 0.821, p = .372$). Table 5 contains effect sizes and confidence intervals for the main effects of time-point and interaction effects. There were no differences in tic severity outcomes found when comparing September to November groups.

[Insert Tables 4 and 5]

Percentage change scores by group are given in Table 6. Figures 2 and 3 show individual participant percentage change in motor tic severity scores between baseline and post-assessment, in the HRT and Education groups respectively.

[Insert Figures 2 & 3]

QoL Outcomes

The repeated measures ANOVA showed the main effect of time-point was on the cusp of significance ($F(1,31) = 4.14, p = .050$, Cohen's $d = 1.14$, large effect) on GTS-QoL Total. Mean (SD) scores improved from 34.60 (2.79) at baseline to 30.24 (2.51) at follow-up. The interaction between group and time-point was non-significant ($F(1,31) = 0.019, p = .892$), as was the main effect of group ($F(1,31) = 0.001, p = .975$). Table 6 shows percentage changes in scores.

[Insert Table 6]

Secondary analyses showed that the main effect of time-point was significant when only including participants who attended five or more sessions, those who had not had concurrent therapy and those who had not experienced stressful life events during the study. Comparing participants who attended the September ($n = 16$) and November groups ($n = 17$) showed that the main effect of time-point on QoL was much stronger in November groups across conditions, although there was no significant difference found at baseline. The effect was non-significant

with a negligible effect size in September groups ($p = .934$, Cohen's $d = 0.006$, negligible effect), but significant with a medium effect size in November groups ($p = .006$, Cohen's $d = 0.474$).

Discussion

This study aimed to compare HRT and Educational groups for children with TS and CTDs in relation to tic severity and QoL. It was hypothesised that the HRT group would experience greater reductions in tic severity compared to the Educational group and that both groups would show significant post-treatment improvements in QoL.

Motor tics, as hypothesised, improved significantly more in the HRT group, representing a medium effect size ($d = 0.55$) and a reduction in score of 14.3%. This is probably because, unlike the educational intervention, HRT focussed directly on reducing tics. Piacentini et al. (2010) reported a medium effect size of $d = 0.68$ following eight individual HRT sessions, with reductions in motor tic severity scores of 27%. The reduction found in the current study is smaller than the 25% considered clinically meaningful by Jeon et al. (2013). This suggests that the effect of group interventions may be diluted compared with individual treatments.

In contrast, the expected improvement in the HRT group was not found on the phonic tic severity subscale, possibly because only 27% of tics the children chose to tackle were phonic tics and therefore the opportunity for change was limited. Previous studies have shown mixed results when comparing phonic and motor tics. Piacentini et al. (2010) found slightly greater effect on phonic tics whereas Wilhelm et al. (2012) found slightly larger effect on motor tics, which may be attributable to the tics selected in therapy.

The QoL effects were somewhat equivocal. In the ITT analysis the main effect of time-point was on the cusp of significance and there was no indication of a group-time interaction. The main effect of time-point showed a large effect size ($d = 1.14$) and improvements in QoL scores of 12.6% across conditions. Analysing only participants who attended five or more

sessions, this effect was more clearly significant, suggesting the intervention dose may be important. Similarly, the effect was significant when analyses excluded participants who had either experienced stressful life events or received concurrent therapy.

Comparing children who attended the groups in September-October with those who attended in November-January revealed a large difference in QoL results. Children in the November-January groups showed a greater response, with large effect, while those in the September-October groups showed negligible effect. There are several possible explanations for this difference. Several children mentioned feeling stressed about returning to school in September (start of the new school year) which may have impacted QoL. Alternatively, despite complete fidelity to the protocols, delivery of both interventions may have improved with experience. In November groups QoL improvements were seen for children in both conditions. Although in the absence of a waiting list control group, the finding is tentative and could be a statistical artefact, it is interesting to consider mechanisms of change given the contrasting interventions. Change may have resulted from an element common to both interventions, such as social support and reduced stigma gained from group therapy. Alternatively it may have been caused by differing mechanisms, such as in the HRT group via increased tic control and in the Education group via improved coping through use of cognitive behavioural strategies. A qualitative investigation of factors influencing QoL in children with TS (Cutler et al., 2009) identified “fitting in with peers” and “attempts to control tics” as important factors. Other important factors were emotional well-being, bullying and physical pain from tics. The two interventions may impact differently on these factors.

In summary, these analyses do not provide conclusive results regarding the relative effects of the interventions on QoL immediately post-intervention. This is consistent with previous research (Woods et al., 2011) showing no difference in psychosocial outcome between HRT and psychosocial support immediately following treatment. However, Woods et al. (2011)

showed greater improvement in the HRT group compared with controls over time, suggesting that consolidation of strategies learnt might lead to secondary improvements in other areas and consequent additional improvements in QoL.

The current study has several important strengths including randomisation to group, blinding of researchers, low attrition and use of ITT analyses. External validity was high due to a clinical sample and few exclusion criteria. The study's low attrition and good attendance rates suggest acceptability of the group interventions. Importantly, tics did not worsen following exposure to others with tics, providing reassurance to families concerned about this (Woods, Conelea, & Himle, 2010).

The study was however relatively underpowered for detecting smaller effects and the ITT analyses may have been overly conservative, resulting in potential for Type II error. Lack of additional control groups limited possible conclusions. Given relatively low acceptance rates among invited families, findings may be generalizable only to children recruited from a single specialist clinic, with sufficient economic resources or practical support to facilitate attendance. Nonetheless, the children showed a range of tic severity and co-morbid symptoms. Given the high prevalence of TS, such an intervention could usefully be made available to larger numbers of children through local clinics if found equally effective in such settings.

If future studies confirm that group-based interventions have a slightly weaker effect than similar length individually-delivered interventions, it would be interesting to increase the number of group sessions to see if this enhances effectiveness. It is noteworthy that many studies of individual HRT have used more than eight sessions (see Verdellen, et al., 2011). If increased effects were demonstrated, the groups would probably still be more cost-effective than individual treatment as large number of patients can be treated at once.

The finding that QoL did not increase for children who returned to school during the intervention has important clinical implications. The school experience in general, transitions to

new schools, or both may impact heavily on the QoL of children with TS, or on their ability to benefit from psychological interventions. The particular challenges faced at these important stages may be a key area for future TS research.

Replication of the study with a larger sample size will be important. Inclusion of individual treatment control groups, would allow conclusions to be drawn about the impact of a group-based format. Future studies could measure potential mediators of change in each intervention, to develop an understanding of the most effective mechanisms of change. It may also be that choice of tic or number of tics treated impacts on change scores. For example, future studies with larger samples could usefully address whether those participants who chose vocals tic for intervention achieved as effective a response, as those who did not choose vocal tics.

Further research is needed to fully determine whether therapy groups are beneficial for children with TS. In the longer term, demonstrating the effectiveness of group therapy would have the potential to increase the number of treatment options available, increase the cost-effectiveness of interventions delivered and reduce waiting times. Studies could specifically address the relative cost-effectiveness of individual and group formats (Tucker & Oei, 2007).

Conclusion

The present RCT was the first to investigate feasibility and efficacy of HRT and Education groups for children with TS. Good attendance in both groups suggested feasibility and acceptability. Results suggest significant tic severity improvements across both groups, with the HRT group showing greater improvements than the Education group.

Modest improvements in QoL were seen in both groups. Group treatments are a promising area for future research in children with TS.

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Tables

Table 1
Details of Tics Chosen to Treat by Participants in the HRT Group

Participant	Tic 1	Tic 2	Tic 3
1	Falling out of chair tic (motor)	Swearing tic (vocal)	None chosen
2	Nose movement (motor)	Record not available	Record not available
3	Head tic (motor)	None chosen	None chosen
4	Eye rolling (motor)	Humming tic (vocal)	None chosen
5	Mouth movement (motor)	Neck rolling (motor)	None chosen
6	Upper body tic (motor)	Eye blinking (motor)	Vocal tic
7	Eye blinking (motor)	Record not available	Record not available
8	Verbal tic (vocal)	Throat clearing (vocal)	None chosen
9	Squeaking tic (vocal)	Verbal tic (vocal)	None chosen
10	Facial movement (motor)	Shoulder and neck tic (motor)	Shoulder tense (motor)
11	Breathe in (vocal)	Jaw click (motor)	None chosen
12	Eye blinking (motor)	Mouth stretch (motor)	None chosen
13	Eye rolling (motor)	Head shake (motor)	None chosen
14	Leg kicking tic (motor)	Upper body rock (motor)	None chosen
15	Shoulder tic (motor)	Chest rub (motor)	Ear swipe (motor)

Table 2
Descriptive Data for Continuous Variables and Group Differences at baseline

	Group			Independent samples <i>t</i> -test <i>p</i>
	All (<i>n</i> = 33)	Education (<i>n</i> = 16)	HRT (<i>n</i> = 17)	
	<i>M</i> (<i>SD</i>)	<i>M</i> (<i>SD</i>)	<i>M</i> (<i>SD</i>)	
Age in years	10.96 (1.45)	11.05 (1.62)	10.87 (1.31)	.73
FSIQ score	101.81 (12.48)	103.13 (13.75)	100.65 (11.55)	.58
YGTSS Motor Severity	17.00 (4.00)	16.31 (3.03)	17.65 (4.74)	.34
YGTSS Phonic Severity	12.67 (6.40)	12.63 (5.93)	12.71 (6.99)	.97
GTS-QoL Total Score	34.61 (15.78)	34.38 (13.76)	34.82 (17.90)	.94

Table 3

Descriptive Data for Categorical Variables and Group Differences at baseline

		Group			Fisher's Exact Test
		All (n = 33)	Education (n = 16)	HRT (n = 17)	<i>p</i>
Gender	Male	25	12	13	1.00
	Female	8	4	4	
Ethnicity	White British	23	12	11	0.71 ^a
	Other White	7	3	4	
	British Indian	1	1	0	
	Black British	1	0	1	
	Mixed/ multiple ethnic	1	0	1	
Tic Disorder	TS	30	15	15	1.00
	CMTD	3	1	2	
ADHD - inattentive symptoms ^b	Clinical level	15	5	10	0.17
	Non-clinical	18	11	7	
ADHD – hyperactive symptoms ^b	Clinical level	5	2	3	1.00
	Non-clinical	28	14	14	
OCD -symptom impairment ^c	Clinical level	11	6	5	0.72
	Non-clinical	22	10	12	
Medication at baseline ^d	Yes	11	4	7	0.47
	No	22	12	10	
Recruitment source	New referral	22	10	12	0.72
	Retrospective	11	6	5	
Month group began	September	16	6	10	0.30
	November	17	10	7	

^a Fisher's exact test conducted comparing British and non-British participants in a 2 x 2 contingency table.^b SNAP IV parent ratings.^c ChOCI-R parent ratings.^d Five clonidine hydrochloride; 2 aripiprazole; 1 atomoxetine, 1 clonazepam, 1 lamotrigine, 1 methylphenidate

Table 4

Means and Standard Deviations by Group and Time-point

		Education (n = 16)		HRT (n = 17)		All (n = 33)	
		Time 1	Time 2	Time 1	Time 2	Time 1	Time 2
YGTSS Motor tic severity	<i>M (SD)</i>	16.31 (3.03)	15.88 (2.28)	17.65 (4.74)	15.12 (4.30)	16.98 (0.70)	15.50 (0.60)
	95% CI	[14.27, 18.36]	[14.11, 17.65]	[15.67, 19.63]	[13.40, 16.84]	[15.56, 18.40]	[14.26, 16.73]
YGTSS Phonic tic severity	<i>M (SD)</i>	12.63 (5.93)	11.13 (5.82)	12.71 (6.99)	12.71 (5.61)	12.67 (1.13)	11.92 (0.99)
	95% CI	[9.31, 15.94]	[8.21, 14.04]	[9.49, 15.92]	[9.88, 15.53]	[10.36, 14.97]	[9.89, 13.94]
GTS-QoL Total Score	<i>M (SD)</i>	34.38 (4.01)	30.31 (3.60)	34.82 (3.89)	30.18 (3.50)	34.60 (2.79)	30.24 (2.51)
	95% CI	[26.20, 42.55]	[22.96, 37.66]	[26.89, 42.75]	[23.05, 37.31]	[28.91, 40.29]	[25.12, 35.36]
		Education (n = 6)		HRT (n = 10)		All (n = 16)	
<i>GTS-QoL Total (Sept Group)</i>	<i>M (SD)</i>	33.17 (14.76)	32.33 (8.48)	32.22 (16.81)	32.56 (16.48)	32.60 (15.48)	32.47 (13.45)
	95% CI	[17.68, 48.65]	[23.44, 41.23]	[19.30, 45.15]	[19.89, 45.22]	[24.03, 41.17]	[25.02, 39.91]
		Education (n = 10)		HRT (n = 7)		All (n = 17)	
<i>GTS-QoL Total (Nov Group)</i>	<i>M (SD)</i>	36.13 (15.42)	28.63 (11.06)	32.50 (19.24)	18.83 (12.97)	34.57 (16.55)	24.43 (12.48)
	95% CI	[23.23, 49.02]	[19.38, 37.87]	[12.31, 52.69]	[5.22, 32.44]	[25.02, 44.13]	[17.22, 31.64]

Table 5
Cohen's d and Mean-Difference Effect Sizes for Time 2 Group Differences and Mean Differences Across Time in Whole Sample

	YGTSS Motor Tic Severity	YGTSS Phonic Tic Severity	GTS-QoL Total
Group Differences at Time 2 (Interaction effect)			
Time 2 mean difference	0.76	-1.58	0.13
Adjusted mean difference ^a	2.1	-1.5	0.57
<i>d</i>	0.22	-0.28	0.04
Adjusted <i>d</i> ^a	0.55	-0.26	0.15
[95% CI]	[-0.16, 1.27]	[-0.97, 0.44]	[-0.56, 0.85]
<i>p</i>	.013	.372	.892
Observed power	.72	.14	.05
Time 1 to Time 2 Differences in Whole Sample (Main Effect of Time)			
Mean difference	1.48	0.75	4.36
<i>d</i>	1.58	0.49	1.14
[95% CI]	[1.13, 2.03]	[0.20, 0.78]	[0.75, 1.53]
<i>p</i>	.001	.372	.050
Observed power	.95	.14	.51

Note. Effect sizes are reported such that for interaction effects positive values indicate greater improvement in the HRT group and improvement in symptoms over time for the main effect of time. Approximate 95% confidence intervals are given (see formulae recommended by Nakagawa and Cuthill, 2007). The variables met necessary test assumptions including absence of univariate outliers and normality.

^aEffect size measures (both mean difference and Cohen's *d*) adjusted for Time 1 group differences on each measure, as recommended by Durlak (2009). Non- adjusted figures are also reported.

Table 6
Percentage Change^a in Scores from Time 1 to Time 2

	YGTSS Motor tic severity	YGTSS Phonic tic severity	GTS-QoL Total
Education (<i>n</i> = 16)	2.6%	10.5%	11.8%
HRT (<i>n</i> = 17)	14.3%	0%	13.3%
All (<i>n</i> = 33)	8.7%	5.9%	12.6%

^aIn all cases these represent reductions in scores in which are associated with reduced tics or improved QoL.

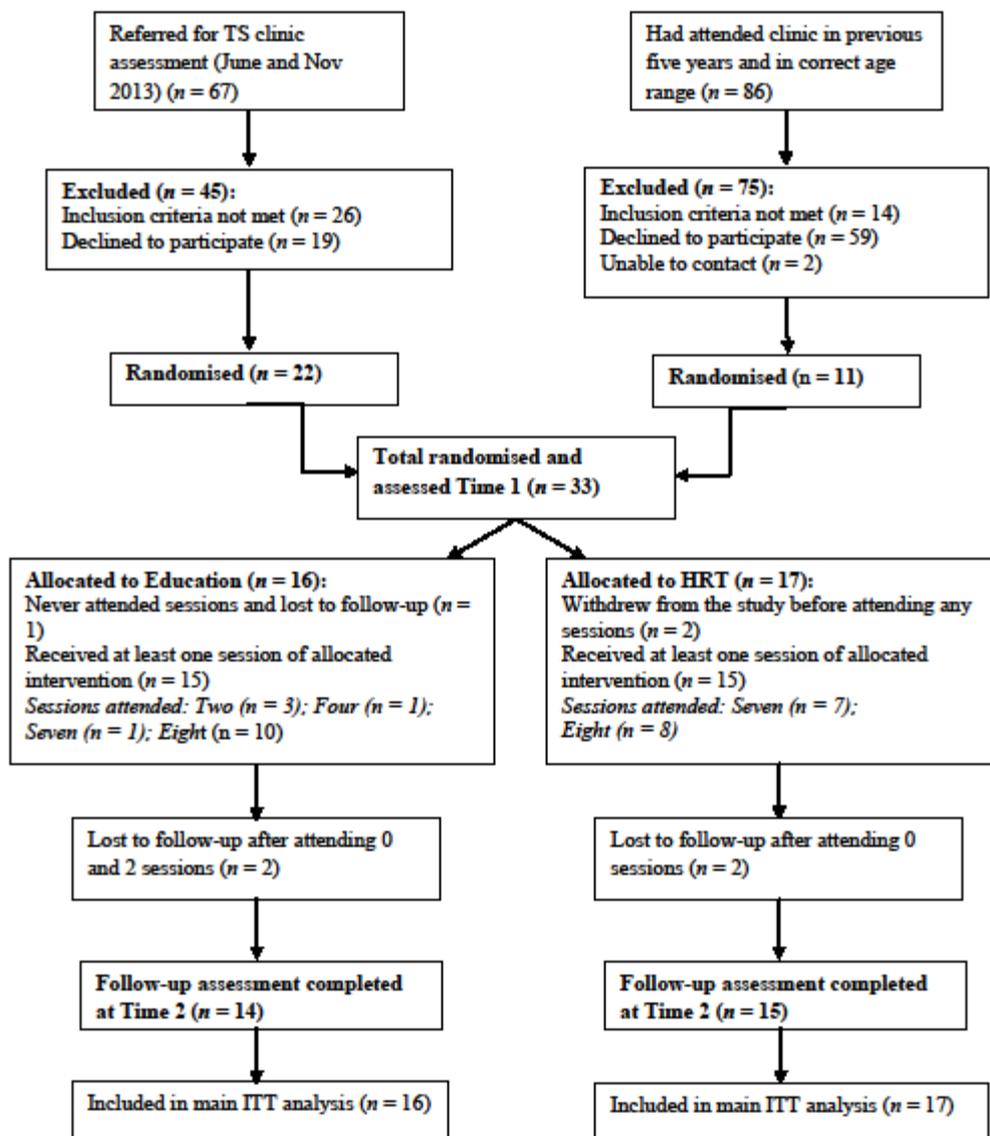


Figure 1. Flow chart of participants through the study.

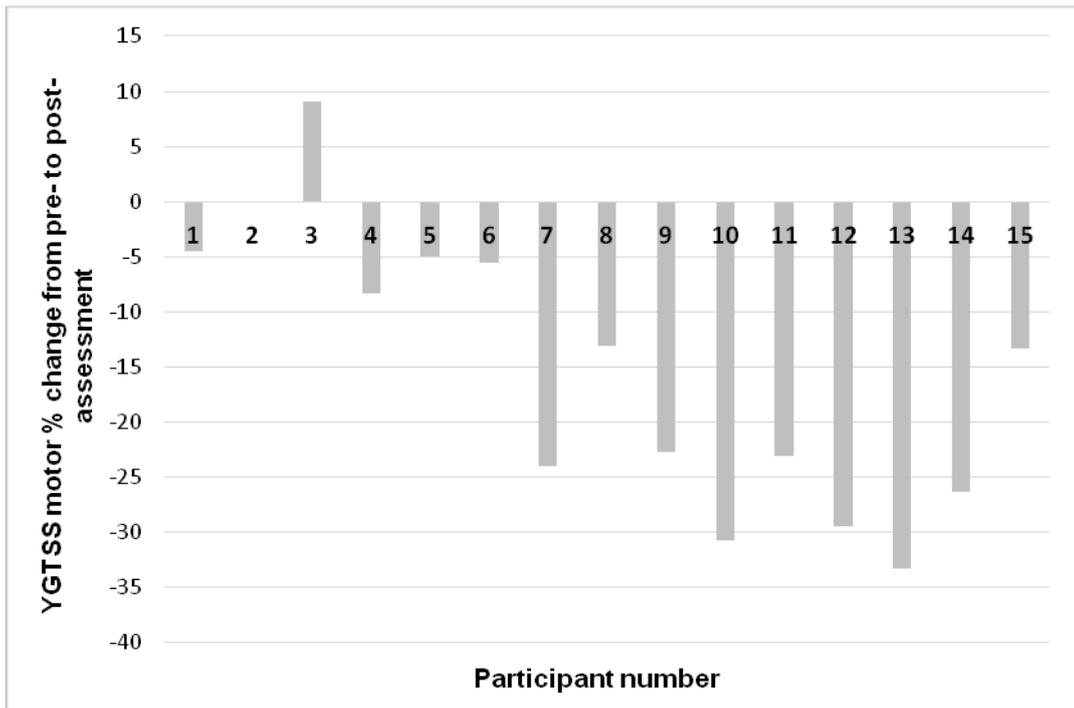


Figure 2: Case-by-case analysis of YGTSS motor tic severity percentage change from pre- to post-assessment (HRT group)

Note. Two participants who did not attend any groups were not included in this analysis.

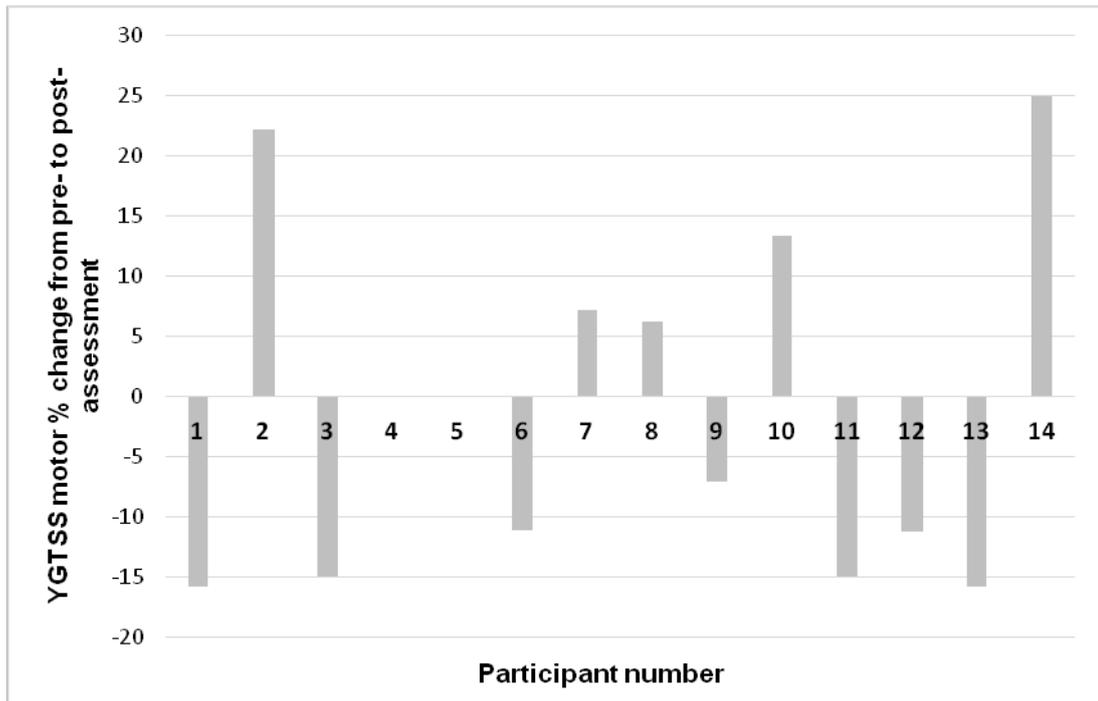


Figure 3: Case-by-case analysis of YGTSS motor tic severity percentage change from pre- to post-assessment (Educational group)

Note. One participant who did not attend sessions, and one lost to follow-up, were excluded from this analysis. Participant #2 had experienced a significant life event. Participant #14 had experienced a change in medication.

References

- American Psychiatric Association. (2013). *The Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition: DSM 5* (Fifth.). Arlington, VA, US: American Psychiatric Publishing Inc.
- Bang, H., Ni, L., & Davis, C. E. (2004). Assessment of blinding in clinical trials. *Controlled Clinical Trials*, 25(2), 143–156.
- Bloch, M. H., & Leckman, J. F. (2009). Clinical course of Tourette syndrome. *Journal of Psychosomatic Research*, 67(6), 497–501.
- 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.
- Crawford, J. R., Anderson, V., Rankin, P. M., & MacDonald, J. (2010). An index-based short-form of the WISC-IV with accompanying analysis of the reliability and abnormality of differences. *British Journal of Clinical Psychology*, 49(2), 235–258.
- 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.
- Durlak, J. A. (2009). How to Select, Calculate, and Interpret Effect Sizes. *Journal of Pediatric Psychology*, 34(9), 917–928.
- Freeman, R. D., Fast, D. K., Burd, L., Kerbeshian, J., Robertson, M. M., & Sandor, P. (2000). An international perspective on Tourette syndrome: selected findings from 3500 individuals in 22 countries. *Developmental Medicine & Child Neurology*, 42(7), 436–447.
- Himle, M., 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.
- 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.
doi:10.1016/j.cct.2013.08.012.
- Leckman, J. F., Riddle, M. A., Hardin, M. T., Ort, S. I., Swartz, K. L., Stevenson, J., & 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, 14–19.
- Lewin, A. B., Murphy, T. K., Storch, E. A., Conelea, C. A., Woods, D. W., Scahill, L. D., ... Walkup, J. T. (2012). A phenomenological investigation of women with Tourette or other chronic tic disorders. *Comprehensive Psychiatry*, 53(5), 525–534.
- 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.
- Murphy, T., & Heyman, I. (2007). Group Work in Young People with Tourette Syndrome. *Child and Adolescent Mental Health*, 12(1), 46–48.
- Nakagawa, S., & Cuthill, I. C. (2007). Effect size, confidence interval and statistical significance: a practical guide for biologists. *Biological Reviews*, 82(4), 591–605.

- 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.
- Piacentini, J. C., Woods, D. W., Scahill, L., Wilhelm, S., Peterson, A. L., Chang, S., ... Walkup, J. T. (2010). Behavior therapy for children with Tourette disorder. *JAMA: The Journal of the American Medical Association, 303*(19), 1929–1937.
- Scharf, J. M., Miller, L. L., Mathews, C. A., & Ben-Shlomo, Y. (2012). Prevalence of Tourette syndrome and chronic tics in the population-based Avon longitudinal study of parents and children cohort. *Journal of the American Academy of Child & Adolescent Psychiatry, 51*(2), 192–201.
- Storch, E. A., Lack, C. W., Simons, L. E., Goodman, W. K., Murphy, T. K., & Geffken, G. R. (2007). A Measure of Functional Impairment in Youth with Tourette's Syndrome. *Journal of Pediatric Psychology, 32*(8), 950–959.
- Sukhodolsky, D. G., Vitulano, L. A., Carroll, D. H., McGuire, J., Leckman, J. F., & Scahill, L. (2009). Randomized Trial of Anger Control Training for Adolescents With Tourette's Syndrome and Disruptive Behavior. *Journal of the American Academy of Child & Adolescent Psychiatry, 48*(4), 413–421.
- Swanson, J. M., Kraemer, H. C., Hinshaw, S. P., Arnold, L. E., Conners, C. K., Abikoff, H. B., Greenhill, L. L. (2001). Clinical relevance of the primary findings of the MTA: success rates based on severity of ADHD and ODD symptoms at the end of treatment. *Journal of the American Academy of Child & Adolescent Psychiatry, 40*(2), 168–179.
- Treasure, T., & MacRae, K. D. (1998). Minimisation: the platinum standard for trials? *BMJ, 317*(7155), 362–363.

- Tucker, M., & Oei, T. P. S. (2007). Is Group More Cost Effective than Individual Cognitive Behaviour Therapy? The Evidence is not Solid Yet. *Behavioural and Cognitive Psychotherapy*, 35(1), 77.
- Uher, R., Heyman, I., Turner, C. M., & Shafran, R. (2008). Self-, parent-report and interview measures of obsessive-compulsive disorder in children and adolescents. *Journal of Anxiety Disorders*, 22(6), 979–990.
- Verdellen, C., Keijsers, G. P. J., Cath, D. C., & Hoogduin, C. A. L. (2004). Exposure with response prevention versus habit reversal in Tourettes's syndrome: a controlled study. *Behaviour Research and Therapy*, 42(5), 501–511.
- Verdellen, C., van de Griendt, J., Hartmann, A., & Murphy, T. (2011). European clinical guidelines for Tourette Syndrome and other tic disorders. Part III: behavioural and psychosocial interventions. *European Child & Adolescent Psychiatry*, 20(4), 197–207.
- Verdellen, C., van de Griendt, J., Kriens, S., & van Oostrum, I. (2011). Tics. Workbook for parents. Boom Publishers, Amsterdam, the Netherlands. Retrieved from http://www.uitgeverijboom.nl/upload/Tics_Workbook_for_parents_EN.pdf
- Wilhelm, S., Peterson, A. L., Piacentini, J. C., Woods, D. W., Deckersbach, T., Sukhodolsky, D. G., ... Scahill, L. (2012). Randomized trial of Behavior Therapy for adults with Tourette Syndrome. *Arch Gen Psychiatry*, 69(8), 795–803.
- Woods, D. W., Conelea, C. A., & Himle, M. (2010). Behavior therapy for Tourette's Disorder: Utilization in a community sample and an emerging area of practice for psychologists. *Professional Psychology: Research and Practice*, 41(6), 518–525.
- Woods, D. W., Piacentini, J. C., Chang, S., Deckersbach, T., Ginsburg, G., Peterson, A., Wilhelm, S. (2008). *Managing Tourette's Syndrome: A behavioural intervention for children and adults*. New York, NY: 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. *Journal of Child Neurology*, 26(7), 858–865.