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A new treatment for children with chronic tic disorders – Resource activation

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ABSTRACT

The aim of this pilot study is to evaluate the efficacy of a resource activation (RA) program as an alternative intervention for children and adolescents with tic disorders. RA interventions focus on the strengths and abilities of the patients. This is expected to improve the patients' perceptions regarding their own resources, which may indirectly result in a reduction of tic symptoms, impairment and comorbid conditions.

A within-subject design with two phases (8 weeks diagnostic; 16 sessions treatment) was analyzed using multilevel modeling ($n = 24$). During the treatment phase, significant reductions of tics were found in clinical rating (Yale Global Tic Severity Scale; YGTSS), parent rating and video observation (motor tics). Moreover, an improvement was shown on most tic-related impairment and subjective burden (SB) ratings. No significant improvement was found regarding comorbid problems and self-esteem. Compared to the preceding diagnostic phase, a significant incremental treatment effect emerged in clinical rating of tic symptoms (YGTSS) and video observation (motor tics). This pilot study provides first hints that RA may represent an effective treatment for reducing tic symptoms, impairment and SB. However, further research is needed in order to establish RA as an effective treatment for tic disorders.

1. Introduction

Tics are involuntarily sudden and rapid movements or vocal expressions, which are often preceded by a premonitory urge (American Psychiatric Association, 2013). The vast majority of patients with tic disorders have comorbid mental disorders, with estimations reaching up to 90% in the case of Tourette syndrome (TS) (Freeman et al., 2000; Hirschtritt et al., 2015; Khalifa and Von Knorring, 2006). The most frequent comorbid disorders are obsessive-compulsive disorder (OCD; 50.0%) and attention-deficit/hyperactivity disorder (ADHD; 54.3%) (Hirschtritt et al., 2015). Additionally, the quality of life (QoL) is lower in children with tics compared to controls (Hesapçıoğlu et al., 2014; Storch et al., 2007b) and functional impairment (FI) is reduced (Conelea et al., 2011; Himle et al., 2007; McGuire et al., 2015b; Storch et al., 2007a). Moreover, a recent study showed higher rates of insecure peer attachment, problems in peer relationships, difficulty in making friends, stigmatization and lower levels of social functioning in youth with TS (O'Hare et al., 2015). Patients with tics often state that they miss out on activities because of physical or emotional problems (Eddy et al., 2011) or due to social stigma and feelings of embarrassment (Elstner et al., 2001). Regarding self-esteem, the findings are inconsistent: While some studies reported lower self-concept or self-esteem in youth with tics (Hanks et al., 2016; Hesapçıoğlu et al., 2014; Khalifa et al., 2010), others did not (Bawden et al., 1998; Edell-Fisher and Motta, 1990). Nonetheless, it was found that patients with TS plus additional comorbid diagnoses had lower self-esteem than did patients with TS only (e.g. Hanks et al., 2016; Silvestri et al., 2017). In their review, Silvestri et al. (2018) concluded that poor self-concept and self-esteem seem to be related more to comorbid disorders than to tic severity. Furthermore, a relation between self-perception and QoL was shown. Children and adolescents with TS or TS plus OCD were found to experience significantly more psychosocial stress than a healthy control group (Lin et al., 2007). Psychosocial stress was predictive of future depressive symptoms and OCD severity. Additionally, psychosocial stress and depression were independent predictors of future tic severity.

Parents of children with TS have been found to have parental stress (Lee et al., 2007; Stewart et al., 2015), higher caregiver burden, lower

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self-esteem (Edell-Fisher and Motta, 1990) and to be at greater risk of mental disorders (Cooper et al., 2003). Patients report that their family members argue more frequently (Eddy et al., 2011). This suggests that in order to reduce parental stress and to support positive family relationships, families should be included in the children's treatment.

The broad spectrum of comorbid disorders, stress, reduced self-esteem, problems in peer relationships, overall FI and reduced QoL of patients with tic disorders may be the main reason why interventions aiming primarily to reduce tic symptoms might fall short.

Besides pharmacological treatment, behavior therapy, particularly habit reversal training (HRT) (Azrin and Nunn, 1973; Piacentini et al., 2010; Woitecki and Döpfner, 2011) and exposure with response prevention (ERP) (Verdellen et al., 2004), are recommended treatments with the primary aim to reduce tic symptoms (Verdellen et al., 2011). In the Comprehensive Behavioral Intervention for Tics (CBIT) study, HRT plus relaxation training, psychoeducation, and a function-based approach was found to be more effective than psychoeducation and supportive therapy (Piacentini et al., 2010).

Despite the proven effectiveness of these interventions, a substantial proportion of patients continue to suffer from tic symptoms, comorbid problems and psychosocial impairment at the end of the interventions (Piacentini et al., 2010; Rizzo et al., 2018; Whittington et al., 2016; Woitecki and Döpfner, 2011, 2012). Moreover, medications can have adverse effects (Rössner et al., 2011; Whittington et al., 2016).

Therefore, there is a strong need to develop and evaluate alternative treatments. One possible approach is to develop interventions targeted at the activation of personal resources rather than problem-focused interventions, which aim directly to reduce tic symptoms. According to Grawe and Grawe-Gerber (1999), resource activation (RA) represents a primary principle of change in psychotherapy in addition to problem-focused interventions. All aspects of the emotional process and entire life situation of a patient can be understood as resources. Hence, resources can be found in all intra-personal (e.g. specific skills, interests, goals) and interpersonal (e.g. supporting family members, friends) strengths and abilities. Specific RA interventions (see 2.5 Treatment) reinforce these strengths and abilities, and therefore focus on the healthy part of the personality instead of focusing on the problems (Gassmann and Grawe, 2004, 2006; Grawe and Grawe-Gerber, 1999). A successful RA supports and improves the positive perception of one`s own resources, thus enabling need-satisfying experiences in terms of pleasure, self-control, positive relationships and self-esteem. It can lead to better overall well-being and improved coping with the disorder. These goals are in accordance with third-wave cognitive behavioral therapy approaches, which focus, for example, on mindfulness, well-being, behavioral activation and relationships (Hayes, 2004; Kahl et al., 2012).

Given that many children with TS experience severe stress, negative peer relationships and negative self-esteem, important therapy goals should be to support positive relationships, positive activities and self-esteem. Moreover, the reduction of stress may also have a positive impact on tic symptoms.

In their Comprehensive Integrated Model, Woods et al. (2007) suggest that while tics emerge because of genetic and/or neurobiological factors, they are influenced by individual internal and external antecedents and consequences. In their review, Conelea and Woods (2008) conclude that stress, anxiety, frustration and tension are often related to an increase in tics. Therefore, the occurrence of tics might be influenced by the positive design of situational constraints and other environmental factors, as well as by reducing stress. In a review of behavioral treatments for TS, Frank and Cavanna (2013) discuss the effectiveness of relaxation therapy, emphasizing that most of the studies are single-case studies or very small-sample studies. The results show improvements in tic symptoms, but the effects are often of short duration. For example, in a study by Bergin et al. (1998), children and adolescents in a relaxation group revealed a greater tic symptom reduction compared to a control group, but this difference was not statistically significant. A recent study (Reese et al., 2015) found mindfulness (meditation) to be effective in reducing tics and impairment in older youth and adults.

The therapy program “Living with Tics” (LWT, 10 weekly sessions) has similar goals to those of the present study (e.g. improving tic-related impairment and psychosocial functioning, increasing QoL). In a randomized control trial conducted in youth, the LWT group ($n = 12$) showed significantly greater improvements in impairment and QoL compared to the waiting group ($n = 12$) (McGuire et al., 2015a). However, in contrast to the present study, the treatment protocol was problem-focused. The present study aims to achieve the mentioned goals without focusing on the tics, and instead by conducting RA exercises. To the best of our knowledge, no previous study has assessed the efficacy of RA treatments for children and adolescents with tic disorders. Some studies have used supportive therapy as a control group treatment (Deckersbach et al., 2006; Piacentini et al., 2010; Wilhelm et al., 2003; Wilhelm et al., 2012). There are some similarities between these control group interventions and our study, as all used psychoeducation, therapists were prohibited from providing advice on tic management strategies, and most studies included problem solving. However, there are also some important differences: In the aforementioned studies, the supportive treatment was basically non-directive in nature, and patients (and their families) were allowed to focus on and talk about the tics, related problems and other issues. In the RA treatment used in the current study, talking about tics and other problems was minimized and the treatment was manualized and highly structured. Therapists guided the patients to focus on positive topics, and exercises were set which required active participation of the patients (see 2.5. Treatment).

Deckersbach et al. (2006) ($n = 15$ adults) formulated similar goals to our own: improving self-esteem, life satisfaction and psychosocial functioning by reducing distress and increasing coping skills. Treatment components included encouragement, reassurance, reframing, clarification and ventilation (Novalis et al., 1993; Pinsker, 1997). While improvements in life satisfaction and functioning were shown, there was no reduction in tic symptoms in the supportive therapy group. The only study to include a supportive therapy group comprising children and adolescents (Piacentini et al., 2010) revealed a reduction of tic symptoms and impairment during the course of the treatment.

The present study investigates the efficacy of a manualized RA treatment program in children and adolescents (Perri et al., 2018b). It is expected that RA exercises will improve the patients' perceptions regarding their own resources, which may indirectly result in a reduction of tic symptoms, impairment and comorbid conditions.

It is suggested that a focus on the patient's own strengths and interests as well as other personal internal and external resources by the patient him/herself and his/her family, and a focus on improving relationships and increasing the rate of positive activities, will lead to (a) an increase in self-esteem and (b) a decrease in tic symptoms, comorbid symptoms, and impairment.

2. Method

2.1. Inclusion criteria

Inclusion criteria were age 8 to 19 years and a diagnosis of TS (F95.2) or a chronic motor/vocal tic disorder (F95.1) according to the ICD-10 criteria (World Health Organization, 1992). Further inclusion criteria were (1) tic symptoms with at least moderate severity (YGTSS Total Tic Score > 13 for TS and > 9 for children with motor or vocal tics only) (cf. Piacentini et al., 2010); (2) IQ > 80; (3) ability and willingness to attend at least 21 weekly outpatient treatment sessions. Moreover, (4) children who received tic medication were able to participate if the dosage had been stable for at least one month and no changes were planned during participation in the study. Finally, (5) tic disorders had to be the primary diagnosis. Exclusion criteria were (1)

comorbid diagnosis of autism or psychosis; (2) parallel continuous behavioral therapy.

Clinical diagnoses for tic disorder and comorbid disorders were based on clinical assessment using the rating scales of a German semi-structured clinical interview based on the diagnostic criteria of DSM-IV and ICD-10, which has been shown to be reliable in German samples (DISYPS-II, Döpfner et al., 2008).

2.2. Study design

The ethics committee of the University Hospital, Cologne approved the study and it was registered on Clinical trials.gov (Identifier: NCT02190370). Patients and their parents provided informed consent prior to inclusion in the study. This analysis forms part of a larger clinical trial aiming to assess the effects of HRT and RA in a combined within- and between-subject design. The present analysis uses a within-subject control group design and provides information about the effects of the treatment compared to a diagnostic period.

Tic symptoms were measured at four assessment points (T0 - T3). T0 occurred at the beginning of an 8-week diagnostic phase, which ended with T1. This was then followed by a 16-week treatment phase. T2 took place after the first half of this treatment phase, i.e. after 8 (with a range of +/-1) weekly patient sessions (50 min.) The second half of the treatment phase, which also comprised 8 (+/-1) weekly patient sessions (50 min.), ended with the final assessment point T3. Additionally, two parent sessions were usually conducted during each treatment phase.

2.3. Statistical analysis

Analyses were conducted using multilevel modeling (MLM) (Goldstein, 2003; Hox, 2002; Raudenbush and Bryk, 2002; Snijders and Bosker, 1999), and piecewise linear growth models were computed (Raudenbush and Bryk, 2002; Singer and Willett, 2003). Two different growth rates were calculated for two different time periods: The diagnostic phase represented the first time period, and changes during this phase (T0 – T1) were covered by the growth rate $\beta_{diagnostic}$. The second time period represented the treatment phase (T1 – T3) and was covered by the growth rate $\beta_{treatment}$. The first main objective of the analysis was to demonstrate that the growth rate $\beta_{treatment}$ (change during treatment) was significant. The second main objective was to demonstrate that the growth rate $\beta_{treatment}$ was significantly larger than the growth rate $\beta_{diagnostic}$ (change during diagnostic phase) as a test for treatment effects. To test $\beta_{treatment}$ against $\beta_{diagnostic}$, contrasts were defined ($\beta_{incremental}$). The intercept of the model was assumed to be random, and for reasons of model identification, the growth rates were fixed. The effect sizes (ES) were calculated using the growth rate multiplied by the measurement time intervals divided by the initial standard deviations (T0). Incomplete cases remain in the analysis in multilevel modeling (Maas and Snijders, 2003). This strategy has been found to be appropriate if missing data are missing at random (Rubin, 1976). With regard to descriptive statistics, missing values were not imputed.

2.4. Outcome measures

The primary outcome measure was the symptom score derived from the parent-rated *Symptom Checklist for Tic Disorders* (SCL-TIC-P) (DISYPS-II, Döpfner et al., 2008). In the first part of the SCL-TIC-P, the presence of different tic symptoms is assessed through 14 items. The frequency of occurrence (“not at all” to "constantly - every few minutes", range 0–4) and intensity of present tics ("very mild" to "severe, irritates others", range 1–4) is assessed for each tic for the last week. By multiplying frequency and intensity ratings for each item and then adding the products and dividing by all given tics (14), a tic symptom score (range 0–16) was calculated. The internal consistency of the SCL-TIC-P has been found to be satisfactory ($\alpha = 0.70$ to $\alpha = 0.79$) (Döpfner and Görtz-Dorten, 2017). Additionally, one item of the SCL-TIC-P assesses the sense of controllability of tic symptoms on a 5-point scale (“1 = very low” to “5 = very high”) and two items assess the SB from the perspective of the parent, one regarding the SB for the child, the other regarding the SB for the parent him/herself (“1 = very low, hardly disturbs” to “5 = extreme”).

The teacher- and the self-rated *Symptom Checklist for Tic Disorders* (SCL-TIC-T, SCL-TIC-S) are structured in the same way as the SCL-TIC-P. The SCL-TIC-S is designed for children from the age of 11 years. In the present study, it was completed by all children with the help of their therapists. The internal consistency of the SCL-TIC-S is not satisfactory ($\alpha = 0.61$ to $\alpha = 0.64$), while the internal consistency of the SCL-TIC-T is satisfactory ($\alpha = 0.74$ to $\alpha = 0.76$; Döpfner and Görtz-Dorten, 2017).

In order to assess tic symptoms observed during diagnostic sessions, a clinical tic observation sheet (SCL-TIC-CO) was designed for the purpose of the present study, on the basis of the 14 items of the SCL-TIC-P and recording the frequency of observed tics (“0 = not observed” to “4 = more than 15 times per therapy session”). The intensity rating is similar to that of the SCL-TIC-P.

The *Yale Global Tic Severity Scale* (YGTSS) is a clinician-rated, semi-structured interview, which begins with a checklist of all tics present during the past week. Motor and vocal tics are rated separately according to five domains (number, frequency, intensity, complexity, and interference; each score ranging from 0 to 5) and summed up to yield a Total Tic Score (range 0–50). In addition, an Overall Impairment Rating is assessed (“none” to “severe”, range: 0–50). Several studies have shown satisfactory validity and reliability of the YGTSS (Leckman et al., 1989; Storch et al., 2005; Walkup et al., 1992).

Direct behavioral observations in the clinical setting were conducted through videos of the patient at the end of every treatment session while the child was playing with the therapist (5 min). Moreover, at every assessment point (T0 - T3), the patient was video-recorded while filling out questionnaires. These videos were randomized with regard to sessions and were analyzed by two trained and blinded raters. The partial-interval method was employed, as it has been shown to correlate highly with event-frequency coding (Himle et al., 2006). Video of counts lasting for five minutes were analyzed, as this duration was found to be reliable and stable (Chappell et al., 1994; Himle et al., 2006). An interval length of 10 s was chosen (Himle et al., 2006); thus, 30 intervals were analyzed using the Programs “CowLog” (Pastell, 2016) and “Time Sampler” (Franke, 2016). Each interval was scored separately for motor and vocal tics (each ranged 0 to 30). Analyses have shown satisfactory interrater reliability and validity (Ruch, 2017; Wälde, 2017).

Emotional and behavioral problems were assessed using the German version of the *Achenbach System of Empirically Based Assessment (ASEBA)*. The parent-rated Child Behavior Checklist (CBCL/6–18R; 113 items) and the self-rated Youth Self Report (YSR/11–18R; 112 items; ≥11 years) were assessed at T0, T1 and T3. The Teacher Report Form (TRF/6–18R; 113 items) was rated at T0 and T3. Internalizing, externalizing and total scores were calculated. Satisfactory internal consistency for the three versions was shown (Döpfner et al., 2014).

The adapted German version (Asendorpf and van Aken, 1993) of the *Self-Perception Profile for Children* (SPPC, Harter, 1985) aims to assess the self-rated multidimensional self-concept. In the present study, a further adaptation was made (Perri et al., 2018a) due to difficulties in understanding the answering format (cf. Van Dongen-Melman *et al.*, 1993). Hence, only the positive reference group was chosen (29 items), and approval had to be rated on a 4-point Likert scale (“0 = not true at all” “3 = true”). Internal consistencies are satisfactory ($\alpha = 0.73$ to $\alpha = 0.91$).

The *Questionnaire for Functional Impairment and Quality of Life of Children and Adolescents with Tic Disorders* comprises a patient version (TiQuaLF-S) and a parent version (TiQuaLF-P) (Volk, 2018). Both versions consist of 46 FI items, which are rated on a 4-point Likert scale (range “0 = not at” all to “3 = especially”) twice: first for difficulties

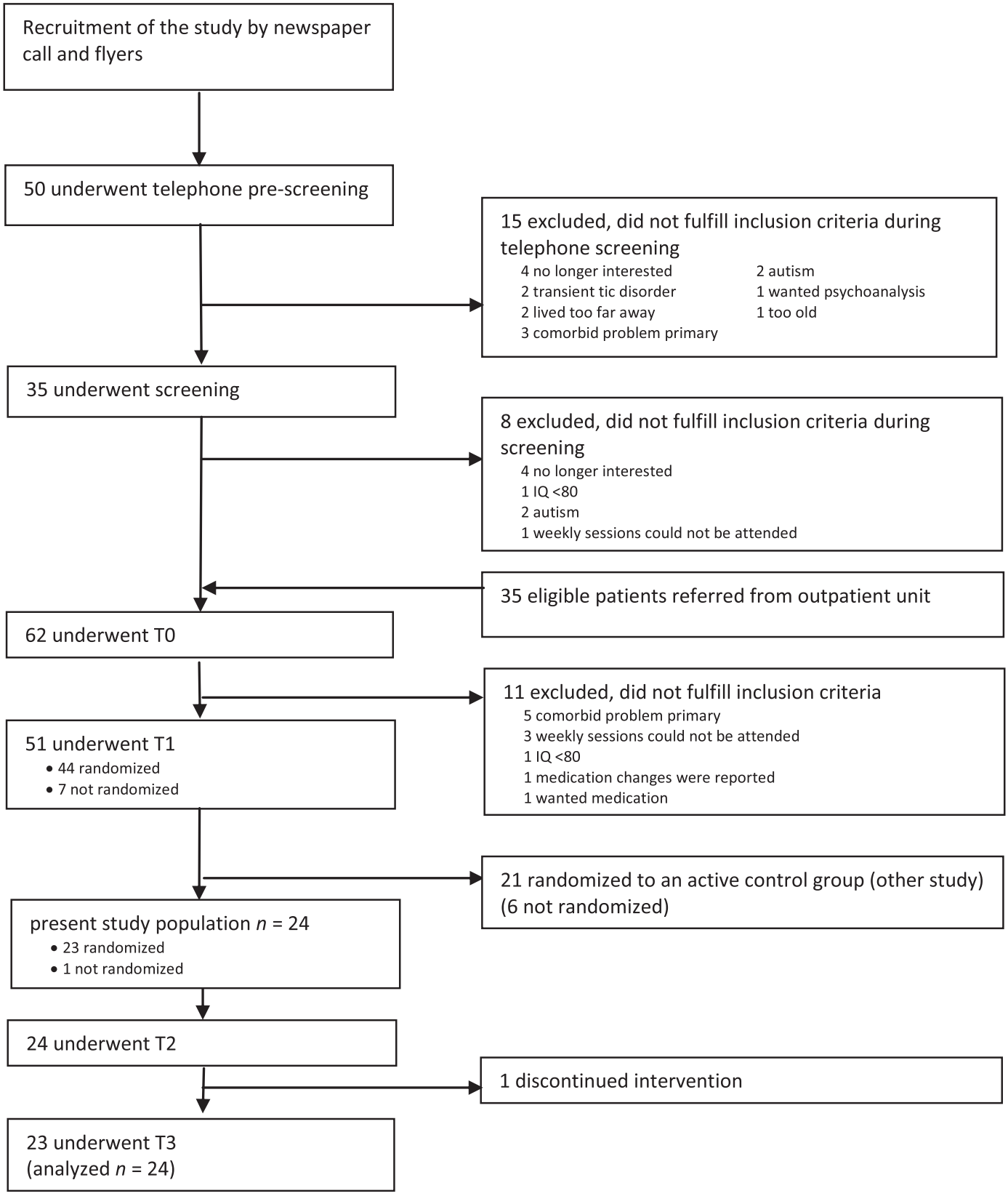

Fig. 1. Flow diagram of the study.

due to tics and second for difficulties caused by other problems. After rating the FI items, the rater assesses the subjective burden (SB) due to tics and other problems. Satisfactory internal consistencies were found for the FI total scales ($\alpha = 0.89$ to $\alpha = 0.95$).

As a *global measure of treatment response*, the Clinical Global Impressions-Improvement scale (CGI-I) (Guy, 1976; Guy and Bonato, 1970) was employed at the final assessment point. A modified version was used, as two ratings were made: tic-specific improvement and

Table 1
Baseline demographic and clinical characteristics.

Characteristic	
Age in years, mean (*SD*) [range]	10;11 (2;6), [8–19]
Male gender; *n* (%)	19 (79.2)
IQ, mean (*SD*), [range]	109.67 (14.64), [84–136]
Diagnosis, *n* (%)	
- Tourette disorder	22 (91.7)
- Chronic motor tic disorder	2 (8.3)
- Chronic vocal tic disorder	0 (0.0)
One or more comorbid diagnoses, *n* (%)	12 (50.0)
- Obsessive-compulsive disorder (F42.X)	3 (12.5)
- Specific (isolated) Phobias (F40.2)	1 (4.2)
- Attention-deficit/hyperactivity disorder (F90.0)	1 (4.2)
- Hyperkinetic Conduct disorders (F90.1)	4 (16.7)
- Separation anxiety disorder of childhood (F93.0)	2 (8.3)
- Adjustment disorders (F42.2)	1 (4.2)
- Other specified behavioral and emotional disorders with onset usually occurring in childhood and adolescence (F98.8)	2 (8.3)
Receiving tic medications at study entry	1 (4.2)
- 100 tiapride (changed temporary (for 2 weeks) during therapy: 100 mg + 50 mg)	1 (4.2)
Receiving other medications at study entry	2 (8.3)
- 36 mg &10 mg methylphenidate (changed to 20 ml lisdexamfetamine + 10 mg methylphenidate; afterwards changed to 20 ml lisdexamfetamine + 0.25 mg risperidone)	1 (4.2)
- 5 mg pipamperone (changed to no medication)	1 (4.2)

improvement in other domains (e.g. self-esteem, comorbid problems). The ratings were discussed and rated in a case conference. Categories ranged from “very much improved” (1) to “very much worse” (7). The CGI-I shows satisfactory validity (Berk et al., 2008).

Treatment satisfaction was measured at the end of the therapy (T3) via the “Therapy Evaluation Questionnaire” (TEQ, Mattejat and Remschmidt, 1998) for patients (≥ 11 years, 20 items) and parents (21 items). Items were rated from 0 (“poor”) to 4 (“excellent”). Total scores and success scores were calculated for both versions. Internal consistencies are satisfactory ($\alpha \geq 0.80$ for most scales) and the retest-reliability for the total score lies between $r = 0.68$ and $r = 0.77$.

2.5. Treatment

The *diagnostic phase* included an intelligence test with the patient as well as a clinical interview with the parents. The treatment phase was based on the manual “Supportive Therapy for Activating Resources in Children” (Perri et al., 2018b) in an individual face-to-face setting. A detailed study plan with concrete exercises for every session was set. These exercises were conducted by the patient with the help of the therapist.

The first treatment phase (T1 to T2) starts with psychoeducation and interventions to strengthen the therapeutic relationship and treatment motivation. Additionally, various exercises are conducted to increase self-esteem, self-confidence and self-acceptance through focusing on the child's strengths, potential and abilities. Example tasks are "My strong name" (for each letter of the patient's name, a strength is written down which begins with this letter) or "My strength tower" (strengths are attached to building blocks). Exercises such as "My strong bonds and supporters" direct the perception towards supporters and positive personal and environmental resources.

The second treatment phase (T2 to T3) combines euthymic, relaxation and mindfulness techniques to enhance well-being in the child. This includes, for example, progressive muscle relaxation, breathing techniques, and mindfulness exercises. In addition, there are exercises to increase well-being, such as the "well-being profile”. Activities with which the patient feels comfortable are highlighted and supported. Furthermore, solution-oriented exercises are conducted which focus on strengths and abilities as key features for achieving individual goals (solution competence). For example, a strong inner figure "superman / superwoman" is imagined. This figure helps the patient to master difficult situations.

Weekly homework is set in order to focus on and implement the strength or learned strategies in daily life. Therefore, a diary is used to note, for example positive situations, the implementation of new strengths, relaxation and mindfulness techniques.

The accompanying involvement of the parents and the entire family serves to consolidate the child's resources, to change the perspective to a positive one, and to establish parental and family resources. For this purpose, for example, reframing exercises are conducted and a “positive time” for the family is implemented.

3. Results

3.1. Participant recruitment

Participant recruitment occurred through a call in a local newspaper and flyers sent to local doctors, psychotherapists and clinicians. In addition, patients were referred from the outpatient unit of the School of Child and Adolescent Cognitive Behavior Therapy at the University Hospital Cologne. After the procedure had been fully explained, all parents and children provided consent to participate in the study. Participant inclusion took place between December 2013 and February 2017. Fig. 1 shows the participant flow of the study.

Of the 51 eligible patients, 24 took part in the present study, while the remaining 27 patients took part in another study on the effects of problem-focused intervention (Viefhaus et al., 2018). One patient discontinued the intervention and dropped out after the first treatment phase (T2) but remained in the analysis. Table 1 shows the demographic and clinical characteristics of the sample. Patients were aged from 8 to 19 years ($M = 10;11$, $SD = 2;6$); 19 (79.2%) were boys, and 22 (91.7%) met the ICD-10 criteria for TS. At study entry, one patient (4.2%) was receiving medication for tics and two patients (8.3%) were receiving medication for comorbid disorders.

3.2. Changes during treatment and treatment effects

Means and standard deviations are shown in Table 2 and results of the MLM analysis are summarized in Table 3.

During the treatment phase ($\beta_{treatment}$), a significant reduction of tic symptoms was shown for the parent-rated SCL-TIC-P (primary outcome), the clinician-rated YGTSS Total Tic Score and the video-observed motor tics in the playing situations. A trend towards a significant improvement during the treatment phase was found for the self-rated

Table 2
Means and standard deviations for all outcome measures assessed at the assessment points T0 to T3.

	T0 (n = 24)		T1 (n = 24)		T2 (n = 24)		T3 (n = 23)	
	AM	*SD*	AM	*SD*	AM	*SD*	AM	*SD*
Tic severity & controllability								
SCL-TIC-P	3.03	1.75	2.50	1.68	1.93	1.55	1.87	1.67
SCL-TIC-S	2.35	1.32	1.85	1.35	2.10	2.18	1.51	1.37
SCL-TIC-T (n_{T0} = 17; n_{T1} = 18; n_{T2} = 15; n_{T3} = 16)	1.47	1.20	1.45	1.61	1.23	1.30	1.49	1.63
SCL-TIC-CO	2.34	1.77	2.20	1.28	2.17	1.70	1.99	1.27
YGTSS- Total Tic Score	23.88	8.92	25.04	9.00	22.38	8.39	20.57	9.26
SCL-TIC-P sense of controllability	2.04	0.81	1.92	0.72	1.83	0.92	1.91	0.95
SCL-TIC-S sense of controllability	2.13	0.99	2.08	0.83	2.21	1.02	2.48	1.24
Video rating motor tics (playing situation)	17.05	6.15	13.26	6.14	16.70	7.73	14.22	7.80
Video rating vocal tics (playing situation)	4.84	5.91	4.78	6.63	3.26	5.94	2.65	4.31
Video rating motor tics (questionnaire situation)	12.14	6.82	16.35	6.98	16.52	7.95	13.14	6.91
Video rating vocal tics (questionnaire situation)	2.50	2.82	3.09	5.02	4.14	6.09	2.68	3.63
Tic-related impairment & subjective burden								
SCL-TIC-P SB child	3.29	1.04	2.79	0.83	2.50	0.98	2.26	1.18
SCL-TIC-P SB parent	3.25	1.03	2.75	0.94	2.50	1.22	2.22	1.04
SCL-TIC-S SB	3.17	1.05	2.96	1.20	2.54	1.25	2.35	1.27
YGTSS Impairment	25.83	10.18	25.42	12.15	19.17	11.77	15.65	10.37
TiQuaLF-P FI tic	0.36	0.27	0.31	0.32			0.26	0.30
TiQuaLF-S FI tic	0.28	0.19	0.19	0.16			0.12	0.15
TiQuaLF-P SB tic	0.82	0.60	0.63	0.55			0.49	0.54
TiQuaLF-S SB tic	0.67	0.58	0.57	0.43			0.25	0.44
Comorbid problems & self-esteem								
CBCL/6–18R Total	30.54	20.71	26.50	20.08			25.52	21.54
CBCL/6–18R internal	8.04	7.37	6.04	4.61			6.04	6.12
CBCL/6–18R external	7.46	6.71	7.38	7.87			7.13	7.62
YSR/11–18R Total (n = 11)	32.73	20.87	21.09	12.59			24.46	16.20
YSR/11–18R internal (n = 11)	11.09	8.69	7.18	6.10			8.55	7.48
YSR/11–18R external (n = 11)	7.46	6.15	4.55	3.39			6.09	5.50
TRF/6–18R Total (n = 15)	32.93	29.73					25.67	25.71
TRF/6–18R internal (n = 15)	6.40	5.25					4.00	4.72
TRF/6–18R external (n = 15)	7.93	10.27					5.73	7.86
TiQuaLF-P FI other problems	0.25	0.29	0.28	0.34			0.25	0.37
TiQuaLF-S FI other problems	0.17	0.20	0.14	0.16			0.12	0.14
TiQuaLF-P SB other problems	0.41	0.53	0.48	0.56			0.49	0.63
TiQuaLF-S SB other problems	0.30	0.40	0.32	0.37			0.19	0.23
SPPC (n_{T0}= 23, n_{T1}= 23, n_{T3}= 22)	2.07	0.55	2.25	0.54			2.20	0.53

Note. n = 24. M = arithmetic mean, *SD* = standard deviation; SCL-TIC-P = parent-rated *Symptom Checklist for Tic Disorders*; SCL-TIC-S = self-rated *Symptom Checklist for Tic Disorders*; SCL-TIC-T = teacher-rated *Symptom Checklist for Tic Disorders*; SCL-TIC-CO = clinical tic observation sheet; YGTSS = *Yale Global Tic Severity Scale*; SB = subjective burden; FI = functional impairment; TiQuaLF-P = parent-rated Questionnaire for Functional Impairment and Quality of Life of Children and Adolescents with Tic Disorders; TiQuaLF-S = self-rated Questionnaire for Functional Impairment and Quality of Life of Children and Adolescents with Tic Disorders; CBCL/6–18R = Child Behavior Checklist 6–18R; YSR/11–18R = Youth Self Report 11–18R; TRF/6–18R =Teacher report Form 6–18R; SPPC = adapted German version of the Self-Perception Profile for Children.

sense of control over tics, and for the video-observed motor tics in the questionnaire situation.

Moreover, significant improvement during treatment was found for SB in TiQuaLF-S and SCL-TIC-P/-S as well as clinician-rated impairment (YGTSS). Improvement with a trend towards significance during the treatment phase was found for self-rated tic-related FI (TiQuaLF-S). Moreover, no significant effects during the treatment phase ($\beta_{treatment}$) were found for comorbid conditions (CBCL/6–18R and YSR/11–18R). The pre- and post-assessment comparison (*t*-test) of TRF/11–18R showed only a trend towards significance for the internalizing scale (p = .072; t (14) = 1.95). Accordingly, no improvement was found for FI and SB ratings regarding other problems ($\beta_{diagnostic}$, $\beta_{treatment}$, $\beta_{incremental}$). No significant effects were found for the SPPC.

Concerning the difference of the growth rates between the treatment phase and the diagnostic phase ($\beta_{incremental}$), a significant treatment effect was only found for the YGTSS and the video-observed motor tics during the questionnaire situation, indicating a greater decrease during the treatment phase compared to the diagnostic phase. No incremental effect was found for the impairment and SB assessments or for comorbid problems and self-esteem ($\beta_{incremental}$).

Regarding the tic-specific improvement (CGI-I), one (4.2%) patient was rated as very much improved, three (12.5%) as much improved and nine (37.5%) as minimally improved. Eight (33.3%) had no change, one (4.2%) was minimally worse, one (4.2%) much worse, and one (4.2%) rating was missing because the patient discontinued the intervention.

Regarding CGI-I rating of the scale “other improvement”, one (4.2%) patient was very much improved, two (8.3%) were much improved, 10 (41.7%) minimally improved, six (25.0%) had no change, three (12.5%) were minimally worse, one (4.2%) was much worse, and one (4.2%) was missing as described above.

Parents rated therapy satisfaction (TEQ; n = 23) at an average of 3.30 (*SD* = 0.48) and success of treatment at 2.77 (*SD* = 0.96). Patients (n = 12) had an average therapy satisfaction of 3.12 (*SD* = 0.57) and rated success of treatment at 2.35 (*SD* = 0.96). A range between 3.5 and 2.5 can be interpreted as "predominantly satisfied" and a range between 2.5 and 1.5 as "partly satisfied".

4. Discussion

During the treatment phase, a significant reduction of tic symptoms with small to medium effect sizes (ES = 0.38 to ES = 0.51) was shown for parent-rated primary outcome (SCL-TIC-P), video-observed motor tics and clinician rating of tic severity (YGTSS), but not for other ratings. However, when comparing pre- and post-assessment (*t*-test) of self-rated tic symptoms, a significant reduction can be found, which might indicate that the reduction is too small to show a significant effect with this sample size in MLM.

On most tic-related FI or SB ratings, an improvement during

Table 3
Results of the multilevel analyses and effect sizes.

	Diagnostic phase (T0–T1)						Treatment phase (T1–T3)						Incremental effect ((T0–T1) -(T1–T3))					
	$\beta_{diag.}$	*SE*	*df*	*t*	*p*	*ES*	$\beta_{treat.}$	*SE*	*df*	*t*	*p*	*ES*	$\beta_{incre.}$	*SE*	*df*	*t*	*p*	*ES*
Tic severity & controllability																		
SCL-TIC-P	−0.61	0.30	69	−2.04	.045	0.35	−0.33	0.16	69	−2.06	.044	0.38	0.29	0.41	69	0.71	.482	0.33
SCL-TIC-S	−0.37	0.27	69	−1.36	.178	0.28	−0.18	0.14	69	−1.24	.218	0.27	0.19	0.36	69	0.52	.605	0.29
SCL-TIC-T (n_{T0} = 17; n_{T1} = 18; n_{T2} = 15; n_{T3} = 16)	−0.05	0.22	45	−0.24	.808	0.04	0.07	0.12	45	0.57	.573	0.12	0.12	0.29	45	0.40	.688	0.20
SCL-TIC-CO	−0.11	0.27	69	−0.41	.684	0.06	−0.12	0.14	69	−0.87	.390	0.14	−0.01	0.37	69	−0.04	.971	0.01
YGTSS- Total Tic Score	1.04	1.18	69	0.88	.382	0.12	−2.27	0.62	69	−3.65	.001	0.51	−3.31	1.59	69	−2.08	.042	0.74
SCL-TIC-P sense of controllability	−0.15	0.16	69	−0.91	.366	0.19	−0.02	0.09	69	−0.21	.833	0.05	0.13	0.22	69	0.59	.557	0.32
SCL-TIC-S sense of controllability	−0.06	0.19	69	−0.34	.738	0.06	0.19	0.10	69	1.90	.061	0.38	0.25	0.25	69	0.99	.325	0.51
Video rating motor tics (playing situation)	−0.02	0.19	415	−0.12	.904	0.03	−0.15	0.06	416	−2.50	.013	0.44	−0.12	0.22	415	−0.57	.570	0.35
Video rating vocal tics (playing situation)	−0.16	0.13	415	−1.25	.214	0.25	−0.01	0.04	416	−0.23	.815	0.04	0.15	0.15	415	1.01	.312	0.52
Video rating motor tics (questionnaire situation)	4.54	1.65	63	2.75	.008	0.67	−1.60	0.86	65	−1.85	.069	0.47	−6.14	2.22	64	−2.77	.007	1.80
Video rating vocal tics (questionnaire situation)	0.77	1.01	63	0.76	.449	0.27	−0.18	0.53	64	−0.34	.738	0.13	−0.95	1.36	63	−0.70	.488	0.67
Tic-related impairment & subjective burden																		
SCL-TIC-P SB child	−0.51	0.20	69	−2.58	.012	0.49	−0.25	0.11	69	−2.40	.019	0.48	0.26	0.27	69	0.97	.337	0.50
SCL-TIC-P SB parent	−0.50	0.21	69	−2.36	.021	0.49	−0.25	0.11	69	−2.26	.027	0.49	0.25	0.29	69	0.86	.393	0.49
SCL-TIC-S SB	−0.25	0.25	69	−0.99	.326	0.24	−0.31	0.13	69	−1.32	.023	0.59	−0.06	0.34	69	−0.18	.861	0.11
YGTSS Impairment	−0.91	1.82	69	−0.50	.619	0.09	−4.78	0.96	69	−4.96	.000	0.94	−3.87	2.46	69	−1.57	.121	0.76
TiQuaLF-P FI tic	−0.06	0.05	45	−1.12	.270	0.22	−0.02	0.03	45	−0.88	.386	0.15	0.04	0.07	45	0.51	.614	0.30
TiQuaLF-S FI tic	−0.09	0.03	45	−2.66	.011	0.47	−0.03	0.02	45	−1.82	.075	0.32	0.06	0.04	44	1.31	.196	0.63
TiQuaLF-P SB tic	−0.18	0.12	45	−1.59	.119	0.30	−0.07	0.06	46	−1.26	.215	0.23	0.11	0.15	45	0.72	.476	0.37
TiQuaLF-S SB tic	−0.10	0.10	45	−0.97	.337	0.17	−0.15	0.05	45	−2.86	.006	0.52	−0.05	0.14	45	−0.36	.721	0.17
Comorbid problems & self-esteem																		
CBCL/6-18R Total	−4.04	2.51	45	−1.61	.114	0.20	−0.60	1.27	45	−0.47	.641	0.06	3.44	3.33	45	1.04	.306	0.33
CBCL/6-18R internal	−2.00	0.85	45	−2.26	.023	0.27	−0.03	0.43	45	−0.07	.948	0.01	1.97	1.13	45	1.75	.086	0.54
CBCL/6-18R external	−0.08	0.91	45	−0.09	.928	0.01	−0.17	0.46	45	−0.37	.717	0.05	−0.09	1.21	45	−0.07	.944	0.03
YSR/11-18R Total (n = 11)	−11.64	6.56	20	−1.77	.091	0.56	1.68	3.28	20	0.51	.614	0.16	13.32	8.68	20	1.54	.140	1.28
YSR/11-18R internal (n = 11)	−3.91	3.01	20	−1.30	.208	0.45	0.68	1.50	20	0.45	.655	0.16	4.59	3.98	20	1.15	.262	1.06
YSR/11-18R external (n = 11)	−2.91	1.65	20	−1.76	.093	0.47	0.77	0.83	20	0.94	.360	0.25	3.68	2.18	20	1.69	.107	1.20
TiQuaLF-P FI other problems	0.03	0.06	44	0.53	.603	0.10	−0.02	0.03	45	−0.67	.509	0.14	−0.05	0.07	44	-0.65	.517	0.35
TiQuaLF-S FI other problems	−0.03	0.04	45	−0.77	.444	0.15	−0.01	0.02	46	−0.29	.773	0.10	0.03	0.05	45	0.47	.640	0.30
TiQuaLF-P SB other problems	0.08	0.12	44	0.63	.530	0.15	0.00	0.06	45	0.04	.968	0.00	−0.07	0.16	45	-0.46	.647	0.26
TiQuaLF-S SB other problems	0.02	0.09	45	0.18	.858	0.05	−0.06	0.05	46	−1.33	.189	0.30	−0.08	0.12	45	-0.65	.522	0.40
SPPC (n_{T0} = 23, n_{T1} = 23, n_{T3} = 22)	0.18	0.12	43	1.58	.122	0.33	−0.03	0.06	43	−0.51	.613	0.11	−0.22	0.16	43	-1.38	.173	0.80

Note. n = 24. β = estimated growth rate; *df* = degrees of freedom; ES = effect size; SCL-TIC-P = parent-rated *Symptom Checklist for Tic Disorders*; SCL-TIC-S = self-rated *Symptom Checklist for Tic Disorders*; SCL-TIC-T = teacher-rated *Symptom Checklist for Tic Disorders*; SCL-TIC-CO = clinical tic observation sheet; YGTSS = *Yale Global Tic Severity Scale*; SB = subjective burden; FI = functional impairment; TiQuaLF-P = parent-rated Questionnaire for Functional Impairment and Quality of Life of Children and Adolescents with Tic Disorders; TiQuaLF-S = self-rated Questionnaire for Functional Impairment and Quality of Life of Children and Adolescents with Tic Disorders; CBCL/6–18R = Child Behavior Checklist 6–18R; YSR/11–18R = Youth Self Report 11–18R; SPPC = adapted German version of the Self-Perception Profile for Children.

treatment was shown on most measures rated by patients and parents (except for TiQuaLF-P) (ES = 0.15 to ES = 0.94). Possibly, patients are more sensitive to changes in their FI and SB compared to their parents. Almost no effects were found for comorbid problems and self-esteem.

A clear treatment effect, as reflected in a significantly greater reduction during the treatment phase compared to the diagnostic phase, was only shown for the YGTSS (ES = 0.74) and the video-observed motor tics in the questionnaire situation (ES = 1.80).

On some questionnaires, a significant improvement was already found during the diagnostic phase. This might be due to some unspecific effects, for instance arising from positive expectations and feeling understood by a specialized therapist (e.g. Frank and Frank, 1991; Grawe and Grawe-Gerber, 1999).

Although a reduction in tic symptoms and impairment was found, it has to be noted that the amount of symptom reduction was lower than that in other studies. The percentage improvement on the YGTSS Total Tic Score of 17.9% is less than that reported in the LWT program (29%) (McGuire et al., 2015a) and the HRT/CBIT studies (30%) (Piacentini et al., 2010; Viefhaus et al., 2018) but almost comparable to that found in the mindfulness study (20.2%, age range 16 to 67) (Reese et al., 2015).

Furthermore, the present study found a reduction of tic-related impairment (YGTSS) of about 39%, which is lower than that found in the LWT program (69.7%) (McGuire et al., 2015a) and the HRT/CBIT studies (about 50%) (Piacentini et al., 2010; Viefhaus et al., 2018), but comparable to that reported in the mindfulness study (38%) (Reese et al., 2015).

Likewise, 16.7% of the patients of the current study improved much or very much during treatment regarding tics (CGI-I). This is comparable to the responder rate of the psychoeducation and supportive group of the CBIT study (18.5%) but is lower than the responder rate in the CBIT group (52.5%) (Piacentini et al., 2010). As Piacentini et al. (2010) found a significant difference between their groups, this suggests that RA is less effective than the CBIT treatment.

Regarding comorbid problems, MLM analyses yielded no significant effects for CBCL/6–18R and YSR/11–18R or FI and SB ratings regarding other problems. This is surprising given that RA is designed to have a broad effect on different problems and it was expected that the intervention mechanism would result indirectly in a reduction of comorbid problems and impairment. However, contrary to expectations, only 50% of the patients had comorbid disorders, compared to comorbidity rates up to 90% for TS found in other studies (Freeman et al., 2000; Hirschtritt et al., 2015; Khalifa and Von Knorring, 2006). Therefore, the present study sample might not be representative regarding comorbid

problems, possibly due to the inclusion criterion that the tic disorder had to be the primary diagnosis. Considering the relatively small sample size of 24 patients in total and 12 patients with a comorbid diagnosis, the sample might be too small to detect possible effects on comorbid problems using MLM. A trend towards significance for internalizing problems was found in the teacher ratings of behavioral and emotional problems (TRF/6–18R). However, further studies with larger sample sizes are needed.

Additionally, the period of treatment time (16 weeks) might have been too short to detect a reduction in comorbid symptoms. It may be that more time is required for the full efficacy of the mechanism of RA to unfold. To evaluate long-term effects, follow-up studies would be necessary.

No significant treatment effect regarding self-esteem was found (SPPC). However, in view of the relatively high positive self-esteem at the start of the treatment (2.25 on a scale ranging from 0 to 3), there was little room left for improvement. Tic patients of this study had a higher mean self-esteem score than patients assessed using the same adapted SPPC with social anxiety disorders (Cremer, 2018) ($M = 1.47$) or oppositional defiant disorders ($M = 1.83$) (Groth, 2018). This is in accordance with some studies which did not find lower self-esteem in tic patients (Bawden et al., 1998; Edell-Fisher and Motta, 1990), although other studies did (Hanks et al., 2016; Hesapçıoğlu et al., 2014; Khalifa et al., 2010). Furthermore, other studies showed that the presence of a comorbid disorder seems to have a greater impact on self-esteem compared to tic severity (Silvestri et al., 2018). As mentioned above, only 50% of the children in this study had a comorbid disorder.

However, although significant effects were not found on all assessments, the clinical impression is that there are some children who improved substantially (cf. CGI-I). This is supported by individual descriptive data of the questionnaires. Additionally, while all patients were offered HRT afterwards, eight of the patients and their parents decided that they did not require further tic treatment.

The clinical impression is that especially younger children, with (sub-) clinical anxieties, low self-esteem and mild to moderate tic severity, benefit from RA. Sukhodolsky et al. (2017) found that anxiety disorders and lower tic severity were predictors of lower tic reduction. Combining these results with the aforementioned clinical impression, this might hint that RA could be a good alternative for children with milder tic severity and comorbid anxieties. Further studies are needed to confirm this hypothesis. Additionally, some patients have negative anticipations of treatments focusing on tics, because they are frightened that tics will increase due to the treatment. Although this has not been found in the long term, for some patients, it might occur during awareness training. Furthermore, some patients find focusing on their tics to be very aversive and confrontational. Another critical aspect of HRT and ERP is motivation. The children and adolescents have to work hard and actively transfer the learned strategies to other settings. As many patients give the feedback that RA “is fun”, it seems to have less of a nature of “hard work”. Therefore, if motivation is lacking, RA might be an alternative or may function as a therapy foundation upon which to build a therapeutic relationship and a therapy motivation for tic-focused treatments.

One of the main limitations of this study is the artificial separation of RA and problem-focused interventions. In real-life therapeutic settings, RA would probably be combined with other treatments such as HRT or Functional Behavior Analyses (e.g. Himle et al., 2014; Woitecki and Döpfner, 2015), which focus directly on the antecedents and consequences of tics.

A further important limitation of the study is that it is not a randomized controlled trial. Advantages of within-subject analyses include the fact that error variance is reduced and fewer participants are needed as patients serve as their own control group (Gliner et al., 2002). Nevertheless, they also bring several disadvantages. It was expected that the course found during the diagnostic phase would continue in a linear fashion during the diagnostic phase. However, particularly during the first weeks after starting a treatment, many studies have reported unspecific treatment effects. Thus, it is unrealistic to assume that this unspecific treatment effect would continue for a further 16 weeks. Accordingly, computing the incremental effect constitutes a very conservative approach, which likely underestimates the treatment effects. Moreover, the small sample size ($n = 24$), with its limited power, may make it difficult to reliably detect treatment effects or differences between the treatment and diagnostic phases.

The large number of outcome variables increases the likelihood of chance findings. However, according to the hypothesis, it was expected that similarly to the tic symptoms, improvement in impairment, self-esteem and comorbidities would also be affected. Therefore, these outcome measurements were correspondingly assessed.

One principal limitation is that in contrast to other studies (e.g. Piacentini et al., 2010), there was no blinded masked clinical rating. To ensure interrater reliability, it would have been beneficial to have an independent and blinded rater rate randomized videos of the YGTSS.

Despite some limitations of this study, the results indicate that RA may be effective in reducing tic symptoms and impairment/SB in some patients with tic disorders. In descriptive terms, the effects seem to be lower than the improvements found in HRT; the clinical impression, however, is that some children benefit very well from RA. Further studies with larger sample sizes are needed to detect predictors of positive treatment outcome (e.g. anxiety, low tic severity) and to confirm the results of this study. Follow-up studies might also detect possible long-term effects.

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Conflict of interest

Manfred Döpfner, Anja Görtz-Dorten and Katrin Woitecki are authors of the treatment manual evaluated and of books about tic disorders or questionnaires used in this study for which they receive royalties from Hogrefe. Other authors have no potential conflict of interests.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.psychres.2019.01.083.

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