

An update on behaviour therapy for Tourette syndrome and chronic tic disorder

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Summary of treatment guidelines

- Psychoeducation is generally recommended as an initial intervention to all patients regardless of symptom severity
- Behaviour therapy (BT) is generally recommended as a first-line treatment
 - Fewer adverse effects compared to medication
 - Two modalities of BT are recommended
 - CBIT/HRT
 - ERP
- Medication is also an evidence-based treatment option

New studies on BT since 2019

- Roughly an update since the 2019 ESSTS Hannover conference
- My selection of studies is not exhaustive
- I have identified a couple of themes

Making treatment more available: Younger population

Development and Open Trial of a Psychosocial Intervention for Young Children With Chronic Tics: The CBIT-JR Study

Shannon M. Bennett*

Weill Cornell Medical College

Matthew Capriotti

San Jose State University

University of California San Francisco

Christopher Bauer

Marquette University

Susanna Chang

UCLA Semel Institute for Neuroscience and Human Behavior

Alex E. Keller

Boston University

John Walkup

Lurie Children's Hospital

- Bennett, S. M., Capriotti, M., Bauer, C., Chang, S., Keller, A. E., Walkup, J., ... & Piacentini, J. (2020). Development and open trial of a psychosocial intervention for young children with chronic tics: The CBIT-JR Study. *Behavior Therapy*, 51(4), 659-669.

Making treatment more available: Younger population



Bennett et al. (2020)

- Aim: Adapt CBIT manual for children younger than 9 years of age, evaluate feasibility, acceptability, and preliminary efficacy
- Design: Open pilot trial
- Sample: N=15; 5-8 years; USA
- Intervention: Some of the adaptations included fewer sessions, larger parent involvement, HRT was delivered through "The Opposite Game"
- Key results: High attendance, high satisfaction, medium within-group effect size ($d=0.73$) on the YGTSS-TTSS, effect maintained at 1-year follow-up

Making treatment more available: Fewer sessions



ORIGINAL RESEARCH: CLINICAL TRIAL



WILEY

Effectiveness of a modified comprehensive behavioral intervention for tics for children and adolescents with tourette's syndrome: A randomized controlled trial

Chia-Wen Chen Doctoral Student¹ | Huei-Shyong Wang MD, Medical Doctor² |
Hsiu-Ju Chang PhD, Professor¹ | Chang-Wei Hsueh MD, Division of Pediatrics, Director³

- Chen, C. W., Wang, H. S., Chang, H. J., & Hsueh, C. W. (2020). Effectiveness of a modified comprehensive behavioral intervention for tics for children and adolescents with tourette's syndrome: A randomized controlled trial. *Journal of Advanced Nursing*, 76(3), 903-915.

Making treatment more available: Fewer sessions



Chen et al. (2020)

- Aim: Limited availability of therapists. Evaluate a shortened version of CBIT (4 sessions instead of 8)
- Design: Single-blind superiority RCT
- Sample: N=46, 6-18 years; Taiwan
- Intervention: CBIT+TAU vs. TAU
- Key results: Small between-group effect ($d=0.44$) on the YGTSS-TTSS. The CBIT-TAU group further improved (in a within-group analysis) between post-treatment and the 3-month follow-up.

Making treatment more available: Intensive + group format



European Child & Adolescent Psychiatry
<https://doi.org/10.1007/s00787-020-01532-5>

ORIGINAL CONTRIBUTION



Tackle your Tics: pilot findings of a brief, intensive group-based exposure therapy program for children with tic disorders

A. P. Heijerman-Holtgreffe^{1,2,3} · C. W. J. Verdellen^{4,5} · J. M. T. M. van de Griendt⁵ · L. P. L. Beljaars^{1,6} · K. J. Kan⁷ · D. Cath^{8,9} · P. J. Hoekstra¹⁰ · C. Huyser¹¹ · E. M. W. J. Utens^{7,11,12}

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- Heijerman-Holtgreffe, A. P., Verdellen, C. W. J., van de Griendt, J. M. T. M., Beljaars, L. P. L., Kan, K. J., Cath, D., ... & Utens, E. (2020). Tackle your Tics: pilot findings of a brief, intensive group-based exposure therapy program for children with tic disorders. *European Child and Adolescent Psychiatry*.

Making treatment more available: Intensive + group format



Heijerman-Holtgreffe et al. (2020)

- Aim: Limited availability of treatment. Evaluate an intensive ERP treatment in a group format, with focus on both tic severity and quality of life outcomes
- Design: Open pilot study
- Sample: N=14, 9-14 years; The Netherlands
- Intervention: "Tackle your tics"-programme: Group-delivered intensive format (same dose as with 12 weekly sessions but delivered over 3 + 1 days) including: ERP, smartphone-app to assist with ERP-practice, coping strategy workshops led by young adult patients, relaxation training, and separate parent meetings.
- Key results: Decrease in YGTSS-TTSS between baseline and the 2-month follow-up ($\eta_p^2=0.41$). Increase in quality of life measured by the C&A GTS-QOL between the same timepoints ($\eta_p^2=0.58$). High treatment satisfaction.

Making treatment more available: Group-format



European Child & Adolescent Psychiatry
<https://doi.org/10.1007/s00787-020-01702-5>

ORIGINAL CONTRIBUTION



Group behavioral interventions for tics and comorbid symptoms in children with chronic tic disorders

Sharon Zimmerman-Brenner^{1,2} · Tammy Pilowsky-Peleg^{3,4} · Lilach Rachamim^{1,5} · Amit Ben-Zvi¹ · Noa Gur^{3,4} · Tara Murphy⁶ · Aviva Fattal-Valevski⁷ · Michael Rotstein^{7,8,9}

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- Zimmerman-Brenner, S., Pilowsky-Peleg, T., Rachamim, L., Ben-Zvi, A., Gur, N., Murphy, T., ... & Rotstein, M. Group behavioral interventions for tics and comorbid symptoms in children with chronic tic disorders. *European Child & Adolescent Psychiatry*, 1-12.

Zimmerman-Brenner et al. (2021)

- Aim: Evaluate CBIT in a group format
- Design: Single-blind superiority (?) RCT
- Sample: N=61, 8-15 years, Israel
- Intervention: Group-delivered CBIT vs. group-delivered education
- Key results: No between-group effect on the primary outcome measure (YGTSS-TTSS?). Between-group effect on the YGTSS Motor Tic Severity Score, as well as the PTQ. Increase in YGTSS Vocal Tic Severity in both groups at post-treatment.

European Child & Adolescent Psychiatry (2019) 28:57–68
<https://doi.org/10.1007/s00787-018-1187-z>

ORIGINAL CONTRIBUTION



Combined habit reversal training and exposure response prevention in a group setting compared to individual training: a randomized controlled clinical trial

Judith B. Nissen^{1,3} · Martin Kaergaard¹ · Lisbeth Laursen¹ · Erik Parner² · Per Hove Thomsen^{1,3}

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Received: 26 February 2018 / Accepted: 23 June 2018 / Published online: 28 June 2018
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- Nissen, J. B., Kaergaard, M., Laursen, L., Parner, E., & Thomsen, P. H. (2019). Combined habit reversal training and exposure response prevention in a group setting compared to individual training: a randomized controlled clinical trial. *European child & adolescent psychiatry*, 28(1), 57-68.

Making treatment more available: Comb. of HRT and ERP, + group format



Nissen et al. (2019)

- Aim: Evaluate combined HRT and ERP, in two delivery formats
- Design: Single-blind open RCT
- Sample: N=59, 9-17 years, Denmark
- Intervention: HRT+ERP, individual vs. group
- Key results: No between-group effect on the primary outcome measure YGTSS (any score). Large within-group effects on the YGTSS-TTSS for both the individual (ES=1.21) and the group format (ES= 1.38).

Making treatment more available: Internet-delivered, with therapist support



European Child & Adolescent Psychiatry
<https://doi.org/10.1007/s00787-020-01686-2>

ORIGINAL CONTRIBUTION



Internet-based guided self-help comprehensive behavioral intervention for tics (ICBIT) for youth with tic disorders: a feasibility and effectiveness study with 6 month-follow-up

Lilach Rachamim^{1,2} · Sharon Zimmerman-Brenner^{1,3} · Osnat Rachamim⁴ · Hila Mualem^{1,2} · Netanel Zingboim¹ · Michael Rotstein⁴

- Rachamim, L., Zimmerman-Brenner, S., Rachamim, O., Mualem, H., Zingboim, N., & Rotstein, M. (2020). Internet-based guided self-help comprehensive behavioral intervention for tics (ICBIT) for youth with tic disorders: a feasibility and effectiveness study with 6 month-follow-up. *European Child & Adolescent Psychiatry*, 1-13.

Rachamim et al. (2020)

- Aim: Evaluate the feasibility and preliminary efficacy of internet-delivered CBIT, with minimal therapist support
- Design: Single-blind (?) RCT
- Sample: N=41, 7-18 years, Israel
- Intervention: Internet-delivered CBIT, with parent- and minimal therapist support, vs. wait-list.
- Key results: Feasible to implement. Large between-group effect ($\eta_p^2=0.20$) on the primary outcome measure YGTSS-TTSS at post-treatment. Participants in the wait-list group were crossed over to CBIT at post-treatment, but a continued large within-group effect ($d=2.25$) was found for the CBIT group at the 6-month follow-up.

Open access

Research

BMJ Open Therapist-guided and parent-guided internet-delivered behaviour therapy for paediatric Tourette's disorder: a pilot randomised controlled trial with long-term follow-up

Per Andrén,^{1,2} Kristina Aspvall,^{1,2} Lorena Fernández de la Cruz,^{1,2} Paulina Wiktor,³ Sofia Romano,³ Erik Andersson,¹ Tara Murphy,^{4,5} Kayoko Isomura,^{1,2} Eva Serlachius,^{1,2} David Mataix-Cols^{1,2}

- Andrén, P., Aspvall, K., de la Cruz, L. F., Wiktor, P., Romano, S., Andersson, E., ... & Mataix-Cols, D. (2019). Therapist-guided and parent-guided internet-delivered behaviour therapy for paediatric Tourette's disorder: a pilot randomised controlled trial with long-term follow-up. *BMJ open*, 9(2), e024685.

Andrén et al. (2019)

- Aim: Evaluate the feasibility, acceptability and preliminary efficacy of internet-delivered HRT and internet-delivered ERP, with minimal therapist support
- Design: Pilot (open) RCT
- Sample: N=23, 7-18 years, Sweden
- Intervention: Internet-delivered HRT and internet-delivered ERP, referred to as "BIP TIC"
- Key results: Feasible and acceptable. Preliminarily efficacious (ERP only). Large within-group effect ($d=1.12$) on the YGTSS-TTSS for the ERP group at the 3-month follow-up. Treatment gains were maintained up to the 12-month follow-up.

Open access

Protocol

BMJ Open Investigating a therapist-guided, parent-assisted remote digital behavioural intervention for tics in children and adolescents – 'Online Remote Behavioural Intervention for Tics' (ORBIT) trial: protocol of an internal pilot study and single-blind randomised controlled trial

Charlotte Lucy Hall,¹ E Bethan Davies,² Per Andrén,³ Tara Murphy,⁴ Sophie Bennett,⁵ Beverley J Brown,² Susan Brown,² Liam Chamberlain,⁶ Michael P Craven,⁷ Amber Evans,⁷ Cristine Glazebrook,⁸ Isobel Heyman,⁹ Rachael Hunter,¹⁰ Rebecca Jones,¹¹ Joseph Kilgariff,¹² Louise Marston,¹³ David Mataix-Cols,¹⁴ Elizabeth Murray,¹⁵ Charlotte Sanderson,¹⁶ Eva Serlachius,¹⁷ Chris Hollis,¹⁸ On behalf of the ORBIT Trial team.

- Hall, C. L., Davies, E. B., Andrén, P., Murphy, T., Bennett, S., Brown, B. J., ... & Hollis, C. (2019). Investigating a therapist-guided, parent-assisted remote digital behavioural intervention for tics in children and adolescents—'Online Remote Behavioural Intervention for Tics' (ORBIT) trial: protocol of an internal pilot study and single-blind randomised controlled trial. *BMJ open*, 9(1).

Hall et al. (2019)

- Aim: Evaluate the efficacy, long-term durability and cost-effectiveness of internet-delivered ERP, with minimal therapist support (BIP TIC)
- Design: Multicentre single-blind superiority RCT
- Sample: N=220, 9-18 years, United Kingdom
- Intervention: Internet-delivered ERP vs. internet-delivered psychoeducation
- Key results: Recruitment is completed but the trial results have not yet been published.

Andréén et al. (2021?)

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Internet-delivered Behaviour Therapy for Children and Adolescents With Tourette's Disorder

⚠ The safety and scientific validity of this study is the responsibility of the study sponsor and investigators. Listing a study does not mean it has been evaluated by the U.S. Federal Government. Know the risks and potential benefits of clinical studies and talk to your health care provider before participating. Read our [disclaimer](#) for details.

ClinicalTrials.gov Identifier: NCT03916055

Recruitment Status: **Recruiting**
First Posted: **April 16, 2019**
Last Update Posted: **September 2, 2020**
[See Contacts and Locations](#)

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- <https://clinicaltrials.gov/ct2/show/NCT03916055>

The ONLINE-TICS Study Protocol: A Randomized Observer-Blind Clinical Trial to Demonstrate the Efficacy and Safety of Internet- Delivered Behavioral Treatment for Adults with Chronic Tic Disorders

*Ewgeni Jakobovski^{1†}, Cornelia Reichert^{1†}, Annika Karch², Nadine Buddensiek¹,
Daniel Breuer³ and Kirsten Müller-Vahl^{1*}*

- Jakobovski, E., Reichert, C., Karch, A., Buddensiek, N., Breuer, D., & Müller-Vahl, K. (2016). The ONLINE-TICS study protocol: a randomized observer-blind clinical trial to demonstrate the efficacy and safety of internet-delivered behavioral treatment for adults with chronic tic disorders. *Frontiers in psychiatry*, 7, 119.

Jakubovski et al. (2016)

- Aim: Evaluate internet-delivered CBIT, without therapist support
- Design: Multicentre single-blind superiority RCT
- Sample: N=166, >18 years, Germany
- Interventions: Internet-delivered CBIT vs. internet-delivered psychoeducation (superiority comparison), plus third smaller group of face-to-face CBIT (feasibility only, not powered for non-inferiority)
- Key results: Recruitment is completed but the trial results have not yet been published.

Optimizations of treatment outcome



www.nature.com/npp

Neuropsychopharmacology



ARTICLE

Optimizing behavior therapy for youth with Tourette's disorder

Joseph F. McGuire¹, Nathaniel Ginder¹, Kesley Ramsey¹, Joey Ka-Yee Essoe¹, Emily J. Ricketts², James T. McCracken² and John Piacentini²

- McGuire, J. F., Ginder, N., Ramsey, K., Essoe, J. K. Y., Ricketts, E. J., McCracken, J. T., & Piacentini, J. (2020). Optimizing behavior therapy for youth with Tourette's disorder. *Neuropsychopharmacology*, 45(12), 2114-2119.

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Optimizations of treatment outcome



McGuire et al. (2020)

- Aim: Augment behaviour therapy with D-cycloserine (DCS)
- Design: Quadruple-blind RCT
- Sample: N=20, 8-17 years, USA
- Intervention: 1 session HRT + 50 mg DCS vs. 1 session HRT + placebo
- Key results: Large between-group effect in favour of HRT + 50 mg DCS ($d = 1.30$).

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Other behavioural treatments: Resource activation



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



Paula Viefhaus^{a,*}, Marion Feldhausen^a, Anja Görtz-Dorten^{a,b}, Helene Volk^a, Manfred Döpfner^{a,b}, Katrin Woitecki^a

^a School of Child and Adolescent Cognitive Behavior Therapy (AKiP) at the University Hospital, Cologne, Pohlstr. 9, Köln D-50969, Germany

^b Department of Child and Adolescent Psychiatry, Psychosomatics and Psychotherapy, Medical Faculty of the University of Cologne, Robert-Koch-Str. 10, Cologne D-50931, Germany

- Viefhaus, P., Feldhausen, M., Görtz-Dorten, A., Volk, H., Döpfner, M., & Woitecki, K. (2019). A new treatment for children with chronic tic disorders–Resource activation. *Psychiatry research*, 273, 662-671.

Other behavioural treatments: Resource activation



Viefhaus et al. (2019)

- Aim: Evaluate the preliminary efficacy of resource activation
- Design: Open pilot study, with 8 weeks baseline phase
- Sample: N=24; 8-19 years, Germany
- Intervention: Resource activation
- Key results: Small to medium within-group effects (ES=0.38-0.51) on the primary outcome (parent-rated SCL-TIC-P), the YGTSS-TTSS and video-observed motor tics, for the treatment phase.

Evidence-based treatment in a naturalistic setting



Child Psychiatry & Human Development
<https://doi.org/10.1007/s10578-020-01098-y>

ORIGINAL ARTICLE



Effectiveness of Behaviour Therapy for Children and Adolescents with Tourette Syndrome and Chronic Tic Disorder in a Naturalistic Setting

Per Andrén^{1,2} · Vera Wachtmeister² · Julia Franzé³ · Caroline Speiner³ · Lorena Fernández de la Cruz^{1,2} · Erik Andersson^{1,2,3} · Elles de Schipper^{1,2} · Daniel Rautio^{1,2} · Maria Silverberg-Mörse² · Eva Serlachius^{1,2} · David Mataix-Cols^{1,2}

- Andrén, P., Wachtmeister, V., Franzé, J., Speiner, C., de la Cruz, L. F., Andersson, E., ... & Mataix-Cols, D. (2020). Effectiveness of Behaviour Therapy for Children and Adolescents with Tourette Syndrome and Chronic Tic Disorder in a Naturalistic Setting. *Child Psychiatry & Human Development*, 1-12.

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Evidence-based treatment in a naturalistic setting



Andrén et al. (2020)

- Aim: Evaluate the effectiveness of BT in a naturalistic clinical setting
- Design: Open naturalistic study
- Sample: N=74, 6-18 years, Sweden
- Intervention: ERP, HRT, or various combinations (including a few cases of concurrent medication), delivered at a tic disorder specialist outpatient clinic
- Key results: Tic severity (YGTSS-TTSS) improved after treatment with a large within-group effect size ($d=1.03$). Tic severity further improved through a 12-month follow-up period.

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Summary and conclusions

- Several identified themes, primarily various ways to make BT more available to patients
 - Reduce costs
 - Reduce the need of trained therapists
 - Reduce the need of travel to specialist clinics
- No new large superiority RCTs have been published, no reason to change current treatment recommendations
- However, 3 large superiority RCTs on internet-delivered behaviour therapy are ongoing, which may have an impact on future treatment recommendations, if shown effective

Other studies published between 2019 and today

Daly, T. (2019). Mobile apps help me manage my tics. *Bmj*, 366.

Khan, K., Hollis, C., Hall, C. L., Davies, E. B., Mataix-Cols, D., Andrén, P., ... & Glazebrook, C. (2020). Protocol for the Process Evaluation of the Online Remote Behavioural Intervention for Tics (ORBIT) randomized controlled trial for children and young people. *Trials*, 21(1), 1-10.

McGuire, J. F., Ricketts, E. J., Scahill, L., Wilhelm, S., Woods, D. W., Piacentini, J., ... & Peterson, A. L. (2020). Effect of behavior therapy for Tourette's disorder on psychiatric symptoms and functioning in adults. *Psychological medicine*, 50(12), 2046-2056.

Nissen, J. B., Parner, E. T., & Thomsen, P. H. (2019). Predictors of therapeutic treatment outcome in adolescent chronic tic disorders. *BJPsych open*, 5(5).

Other studies published between 2019 and today

Petruo, V., Bodmer, B., Bluschke, A., Münchau, A., Roessner, V., & Beste, C. (2020). Comprehensive Behavioral Intervention for Tics reduces perception-action binding during inhibitory control in Gilles de la Tourette syndrome. *Scientific reports*, 10(1), 1-8.

Schaich, A. M., Brandt, V. C., Senft, A., Schiemenz, C., Klein, J. P., Fassbinder, E., ... & Alvarez-Fischer, D. (2020). Treatment of Tourette Syndrome with Attention Training Technique—A case series. *Frontiers in psychiatry*, 11, 936.

Singer, H. S., McDermont, S., Ferenc, L., Specht, M., & Mahone, E. M. (2020). Efficacy of Parent-Delivered, Home-Based Therapy for Tics. *Pediatric Neurology*.

Tilling, F., & Cavanna, A. E. (2019). Relaxation therapy as a treatment for tics in patients with Tourette syndrome: a systematic literature review. *Neurological Sciences*, 1-7.

Wolicki, S. B., Bitsko, R. H., Holbrook, J. R., Danielson, M. L., Zablotzky, B., Scahill, L., ... & Mink, J. W. (2020). Treatment use among children with Tourette syndrome living in the United States, 2014. *Psychiatry Research*, 293, 113400.

Thank you very much!

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