

TOURETTE SYNDROME and BEHAVIOURAL THERAPIES

Behavioural therapies (BT) in Tourette syndrome (TS) broadly focus on quality of life, social stigma, and tics, and are based on the belief that tics are biologically driven behaviours that are influenced by cues. Cues can be external (*activities*, such as homework; and *environmental*, such as being at shop), and internal (premonitory urges, or internal mood states).

Within Tourette Syndrome (TS) tics are brief, sudden movements or sounds, that occur at an inappropriate moment, have variability in severity, suppressibility, and comprise the presence of an anticipatory uncomfortable sensory sensation (premonitory urge). Within BT these habitual responses are viewed as being strengthened through negative reinforcement. Some clinical procedures and therapy manuals are focused on training a patient to recognise the premonitory urge and generate voluntary competing responses incompatible with the tic, known as habit reversal training (HRT); or increase their tolerance to premonitory urge, known as exposure with response prevention (ERP). Both are foundational elements in the primary BT within TS of comprehensive behavioural intervention for tics (CBIT).

Comprehensive Behavioural Intervention for Tics

Over the last decade there has been a resurgence of interest in CBIT due to two, high quality randomised control trials, that identified a decrease in tic severity, with no serious side effects (Piacentini, et al., 2010; Wilhelm, et al, 2012). Research suggests, within CBIT that there is little improvement for patients concurrently taking tic medication (Sukohdolsky, et al., 2017). Improvement in tic suppression within CBIT suggests an association with improved quality of life, and a patient's sense of control over their tics (Dabrowski, et al., 2017). Some CBIT research has focused on tic phenotype, and found that tic phenotype may influence CBIT outcome. Chiu, et al., (2020) found that darting eyes, facial grimaces, head forward, throwing

head back, shrugging shoulders, or quick flexion/extension of arms reported significant improvements, suggesting tic phenotype may influence CBIT outcome. Petruo, et al. (2020) connected CBIT to a new and emerging concept within TS, and provided an insight into why CBIT is effective. Petruo, et al. (2020) suggest that CBIT reduces the 'binding' between perception and action, and increases the ability to perform response inhibition.

CBIT builds on different psychoeducational elements such as relaxation training, identifying situational factors, the development of strategies to reduce the influence of these behaviours, and reward contingency plans. Bennett, et al., (2019) in their family-based adaptation for children under nine years, reported a 100% retention rate at both three months and one year follow up, together with a reduction of tic and related comorbid symptoms, and changes in parental accommodation and attention to tics. Overall, CBIT appears to be an acceptable intervention for individuals with tics that lead to psychosocial or physical impairment, and who are motivated to participate in treatment (Pringsheim, et al., 2019).

Habit Reversal Training

As mentioned above, HRT builds the basis for CBIT and is a primary component of CBIT. HRT is focused around awareness training and an emerging view that tics may represent prefabricated actions stored in event files, which are triggered by appropriate perceptual input. HRT seeks to replace the tic with another response, and endeavours to train the structure to unbind tic-specific event files. Herein the self-monitoring of tics, the premonitory urge associated with them, and the competing response behaviour, makes the voluntary behaviour physically incompatible with the tic when the urge occurs. Within HRT Seragni, et al., (2015) reported an improvement in tic reduction and global functioning, however found no significant changes in patient's quality of life.

Exposure and Response Prevention

ERP aims to interrupt the association between the premonitory urge and tic response in order to prevent the tic from occurring. ‘Exposure’ is exposure to the situation, and ‘response prevention’ is the making of a choice not to do the compulsive behaviour once it is triggered. Some studies suggest that ERP is as effective as HRT in tic reduction (Hoogduin, et al., 2008) although there is currently insufficient evidence (Pringsheim, et al., 2019). Other studies report shorter exposure sessions, as not being inferior to that of prolonged exposure sessions, suggesting that shorter exposure might be not only efficient, but also enable more patients to be reached (van de Griendt, et al., 2017). Research on group and individual settings similarly report no difference, with reductions comparable to other studies, and increased post treatment experience of control (Nissen, et al., 2019).

Psychoeducation and Supportive Therapy (PST)

Patients with tics receiving CBIT are more likely than those receiving PST to have reduced tic severity. Psychoeducation primarily seeks to protect patients from misleading information, and is important for patients, families, and schools. Educational interventions appear to have a greater impact when directed at parents, whom are more likely to influence key phases in a child’s development of self-esteem and personality. Effectiveness of PST has also been found to be dependent upon an active commitment and involvement from a patient’s parents. Challenges do exist within families around the complex dualism of involuntariness and voluntary suppression, that leads to blaming attitudes of some parents towards a child; or high emotional responses, that lead to blame or guilt between parents, or other siblings. Gousse et al., (2016) advocate the ‘matter of fact’ attitude as being the most beneficial and useful. Parent training programs and social support remain important objectives within PST.

PST also focuses on the evaluation of stigma, the ability of the individual to fulfil their aspirations, discrimination, and victimisation at school and work. There is a dearth of research on standardisation in this area, and even less clear research on psychoeducation in adult TS patients. PST has been found to be effective in 9 to 17-year olds and those with co-morbid OCD and ADHD. Children under 10 years may not yet have the awareness around premonitory urges, nor be able to understand and apply strategies. As this is a fairly new treatment rigorous training in its application is sparse. There is a case study of a 10-year-old boy “Hiro” that found that the creation of an associated treatment plan, the creation of an individualised case formulation, and the importance of a therapeutic bond being key factors in effectiveness (Lichtman, 2015).

Accessibility

Although CBIT is the first line of behavioural treatment in TS, it is not widely available. Undertaking an 8 to 10-week course may be impeded by not having a provider nearby, or an inability to attend sessions on a weekly basis. To support ease of accessibility internet based and telehealth approaches continue to evolve. Internet delivered BT also has the potential to greatly increase access to evidence-based treatment for young people (Andren, et al., 2020). Within HRT there is insufficient evidence to determine the relative efficacy of HRT by video conferencing compared to face to face HRT (Pringsheim, et al., 2019).

Technology is also evolving and holds promise for CBIT dissemination where services might not be available. The novel TicHelper (www.tichelper.com) is an online treatment program for youth. TicHelper is interactive, easily navigable, and developmentally tailored approach, although no outcome research on it has been published yet, and TicHelper does not however address comorbidities or tic related psychosocial challenges and is focused primarily on tic reduction (Conelea & Wellen, 2017).

References

- Andren, P., Aspvall, K., Fernandez de la Cruz, L., Wiktor, P., Romano, S., Andersson, E., Murphy, T., Isomura, K., Serlachius, E., & Mataix-Cols, D., (2020). Therapist guided and parent guided internet delivered behaviour therapy for paediatric Tourette's disorder: A pilot randomised controlled trial with long term follow up. *Mental Health Research*.
- Bennett, S.M., Capriotti, M., Bauer, C., Change, S., Keller, A.E., Wallup, J., Woods, D., & Piacentini, J. (2019). Development and open trial of a psychosocial intervention for young children with chronic tics: The CBIT-JR study. *Behaviour Therapy*.
doi:10.1016/j.beth.2019.10.004
- Chiu, S., Deeb, W., Almeida, L., Simpson, H., Okun, M., & Malaty, I. (2020). CBIT response in clinical practice: The potential importance of comorbidities and tic location. *Neurology*, 94(15)
- Conelea, C.A., & Wellen, B.C.M. (2017). Tic treatment gone tech: A review of TicHelper.com. *Cognitive & Behavioural Practice*, 24(3), 374-381.
- Dabrowski, J., King, J., Edwards, K., Yates, R., Heymore, I., Zimmerman-Brenner, S., & Murphy. (2017). 81 group interventions for children with Tourette syndrome: A 12 month follow up study of a randomised controlled trial comparing comprehensive behavioural intervention and psycho-education. www.adc.bmj.com
- Gousse, V., Czernecki, V., Denis, P., et al. (2016). Impact of perceived stress anxiety, depression and social support on coping strategies of parents have a child with Gilles del la Tourette Syndrome. *Archive of Psychiatric Nursing*, 30, 109-113

- Hoogduin, C.A.L., Kato, B.S., Keijsers, G.P.J., Cath, D.C., & Hoijtink, H.B. (2008). Habituation of premonitory sensations during Exposure and Response Prevention in Tourette syndrome. *Behaviour Modification*, *32*, 215-227
- Lichtman, J.D. (2015). The case of 'Hiro': Treating Tourette syndrome by comprehensive behavioural intervention for tics (CBIT). *Pragmatic Case Studies in Psychotherapy*, *13*(1), 1-37
- Nissen, J.B., Kaergaard, M., Larsen, L., Parner, E., & Thomsen, P.H. (2019). Combined HRT and ERP in group setting compared to individual training: A randomised controlled clinical trial. *European Child & Adolescent Psychiatry*, *28*, 57-68
- Petruo, V., Bodmer, B., Bluschke, A., Munchau, A., Roessner, V., & Beste, C. (2020). Comprehensive behavioural intervention for tics reduction perception-action binding during inhibitory control in Gilles de la Tourette syndrome. *Scientific Reports*, *10*, 1174
- Piacentini, J., Woods, D.W., Scahill, L., et al. (2010). Behaviour therapy for children with Tourette disorder: A randomised controlled trial. *JAMA*, *303*(19), 1929-1937
- Pringsheim, T.M., Okun, M.S., Muller-Vahl, K., Martino, D., Jankovic, J., Cavanna, A.E., Woods, D.W., Robinson, M., Jarvie, E., Roessner, V., Oskoui, M., Holler-Managan, Y.K., & Piacentini, J. (2019). Practice guidelines recommendations summary: Treatment of tics in people with Tourette syndrome and chronic tic disorders. *American Academy of Neurology*, *92*, 896-906
- Seragni, G., Chiappedi, M., Bettinardi, B., Zibordi, F., Colombo, T. Reina, C., & Angelini, L. (2015). Habit reversal training in children and adolescents with chronic tic disorders: an Italian randomised single blind pilot study. *Minerva Pediatrica*, *70*(5), 5-11

- Sukhodolsky, D.G., Woods, D.W., Piacentini, J., Wilhelm, S., Peterson, A.L., Katsovich, L., Dziura, J., Walkup, J.T., & Scahill, L. (2017). Moderators and predictors of responses to behavioural therapy for tics in Tourette Syndrome. *Neurology*, 88(11)
- Van de Griendt, J.M.T.M., van Dijk, M.K., Vedellen, C.W.J., & Verbraak, J.P.M. (2017). The effect of shorter exposure versus prolonged exposure on treatment outcomes in Tourette syndrome and chronic tic disorders: An open trial. *International Journal of Psychiatry in Clinical Practice*, 22(4), 262-267
- Wilhelm, S., Peterson, A.L., Piacentini, J., et al. (2012). Randomised trial of behavioural therapy for adults with Tourette syndrome. *Archive of General Psychiatry*, 69, 795-803