REVIEW



Behavioral and psychological

therapies for Tourette syndrome and tic disorders: their place in treatment and history

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Practice points

- Tics are very common in childhood and the majority do not need treatment.
- Most tics tend to 'wax and wane' and a detailed history will elicit the core characteristic features.
- Look for associated comorbid conditions such as obsessive–compulsive disorder and attention deficit disorder.
- If the tics are painful or cause significant psychosocial stress, then consider treatment.
- There is no cure for tics and it is important that the patient and family have this information to avoid unrealistic expectations.
- There are a number of medicines that have been shown to be effective in reducing the tic severity and this is the current treatment of choice.
- Recent studies have shown that a psychological therapy known as habit reversal therapy may be beneficial in reducing tics but this needs to be administered by a trained therapist.

SUMMARY In the past, only psychological therapies were used for Tourette syndrome (TS), thereafter medications were the mainstay for many years. There has been a recent increase in the literature on evidence-based behavioral therapies for TS, but there seems to be little evidence-based treatment of psychological therapies for tic disorders. This article reviews the management of TS, highlighting the controlled studies examining behavioral and cognitive therapies for TS and tic disorders. There have been numerous case reports and small case series on the subject of cognitive-behavioral treatments of TS and tic disorders. There have been ten published studies that used controls as part of the design. There is evidence that habit reversal training and exposure response prevention therapy may have some effect on reducing tics, but, to date, long-term follow-up has not been established. Recent studies using habit reversal training, in particular, have been promising. There are no evidence-based studies showing that other forms of psychological therapies such as psychoanalysis are effective. Further clinical research is needed to establish the effectiveness of several behavioral therapies. Developmental considerations should be considered when looking at its application to children. Suggestions for future studies are discussed.

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Tics are usually transient in childhood and are generally regarded as part of normal development. However, some children go on to develop persistent isolated tics, which may cause social embarrassment whilst other children develop several motor and vocal tics, which can cause significant impairment to the child and family's psychosocial functioning [1,2]. Tourette syndrome (TS) is a common tic disorder (see later) and the tic disorder into which most research efforts are directed. Current guidelines and expert reviews on tic disorders suggest pharmacological intervention as the main component of a management package [2-4]. There is, however, relatively little discussion on nonpharmacological treatments. Given that many of the drugs used often have side effects and that none have been proven to be entirely effective for all individuals [5], and have idiosyncratic effects in different people [3], we feel it important to review all available evidence, in particular the evidence-based literature on nonpharmacological treatments for TS and tic disorders.

This article is a review of nonpharmacological treatments for tics, and in particular TS, and a more indepth review of published controlled trials of psychological/behavioral treatments for TS and/or tic disorders. The literature search for this review involved computerized searches of current OVID/PubMed databases and a review of references from identified studies, with appropriate cross-referencing. We have included all studies (irrespective of patients' age) in this review and attempt to make reference to particular developmental aspects.

Description of the disorders Tics

A tic is a rapid, recurrent, nonrhythmic motor or vocal action. The tic is sudden and purposeless. Motor tics include eye blinking, grimacing and shoulder shrugging. Vocal (often referred to as phonic) tics include coughing, barking, or clearing one's throat. Tics may be simple (isolated, involving only one group of muscles, single or repetitive [blinking]) or complex (co-ordinated, sequential movements resembling normal motor acts/gestures but are inappropriately intense and timed (e.g., touching) and may be repetitive (stereotypic). Tics fluctuate (wax/wane), are suppressible, suggestible and persist during sleep [6].

According to the Diagnostic and Statistical Manual of Mental Disorders [7], tic disorders (TD) can be divided into transient tics, chronic motor or vocal tics and TS.

Transient tic disorders

In these disorders the tics (motor and/or vocal or both) only last a few weeks or months, but for this diagnosis to be made, the tics last for less than a year.

Chronic motor or vocal tics

In these disorders, namely chronic motor tic disorder or chronic vocal tic disorder, the patient will have either motor or vocal but not both and the tics occur for more than a year.

TS (combined chronic multiple motor & vocal tics)

Tourette syndrome is the most common cause of tics. Diagnostic criteria for TS include multiple motor tics and one or more vocal/phonic tics, lasting more than a year [7,8].

The age at onset of TS ranges from 2–21 years, with a mean of 5-7 years being common. The onset of vocal tics is usually later (11 years). Premonitory sensations occur in 80% of patients (children often less able to describe them): they may be localized (around the area of the tic, [e.g., the uneasiness before a sneeze]) or generalized (covering a wide area of the body). Simple vocalizations include sniffing and throat clearing, whilst complex vocal/phonic tics include the use of certain words or phrases. Other symptoms include echolalia (copying what others say), echopraxia (a complex motor tic involving copying what others do) and palilalia (repeating one's last word or part of sentence). Coprolalia (inappropriate, involuntary, swearing) is uncommon, occurring in approximately 10-15% of adult clinic patients, often starting at 11 years of age: it differs from 'social swearing'. Many doctors are under the misapprehension that coprolalia must be present for the diagnosis. Instead of the whole swear word, many say only parts of the word, and also disguise it by coughing. For reviews of the clinical phenomenology see [9-19].

The clinical characteristics of TS are similar irrespective of the country of origin, highlighting the biological nature of TS. In some instances it seems that within families, the affected males have tic symptoms, whereas the females have obsessive-compulsive behavior, symptoms not meeting criteria for severe obsessive-compulsive disorder (OCD) but many of the traits and symptoms [3].

Prior to the onset of the tic, the majority of individuals (particularly older children and adults) often describe a sensation, known as

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premonitory sensory phenomena (PSP) in the region of the tic [20]. The sensation is temporarily relieved after the tic has been discharged. Examples of these sensations include a burning feeling in the eye prior to an eye blink, itching before a movement of the shoulders, or tension in the neck that is relieved by stretching the neck. It is not clear whether younger children experience PSP but are unable to describe it or that the symptom is absent altogether.

Tics usually fluctuate in severity and frequency during the day as well as between days. This is often referred to as 'waxing and waning'. It used to be thought that tics were entirely involuntary but many reports have suggested some degree of voluntary control over tics [21–23].

Comorbidities associated with TS

By far the majority of individuals with TS (90%) also have comorbid disorders, which have been demonstrated in both clinical cohorts [24] and also in community settings [25,26]. The most common comorbid disorders are attention deficit hyperactivity disorder and OCD, both said to occur in approximately 50% of clinical cases. Other comorbid conditions include depression, oppositional defiant disorder, conduct disorder, autistic spectrum disorders and personality disorders [3].

Epidemiology

Approximately 10% of children have had a transient tic at some point before the age of 10 years [3]. TS was once considered to be uncommon, even rare, as suggested by older studies (e.g., identifying TS patients who had been admitted to hospital or were already known to doctors), but recent studies have suggested a prevalence of 1% of young people between the ages of 5 and 18 years [27,28]. Of importance is that these studies were worldwide (Sweden, UK, USA, Poland, Taiwan, Italy and China), and they were similar as they were conducted in mainstream schools, used similar multistaged methods, with both observations and questionnaires about pupils, as well as obtaining information from parents and/or teachers, or both. In addition, they were initiated and conducted by clinicians with a special interest in TS. The majority of the TS 'cases' identified, were undiagnosed and mild, without distress, impairment or coprolalia [27,28]. The prevalence of TS in special educational populations, such as those individuals with emotional and behavioral and/or learning difficulties or autistic spectrum

disorder is higher [27,28]. Robertson suggests that although less obvious/severe, the prevalence in adults is also 1% [27]. Gender differences have been examined on numerous occasions and have showed that TS is more prevalent in boys than girls [1,3–4,14,15,29].

Etiology

Neurobiological models

Although the precise etiology is unknown, several studies support the fact that TS and tic disorders are an inherited, developmental disorder involving an imbalance in the dopamine pathways in the brain, most likely involving the basal ganglia [30-33]. A possible infectious origin for TS has also been identified recently linking the sudden onset of obsessive—compulsive symptoms and tics with sore throat infections caused by group A hemolytic streptococcal bacteria [34]. Other etiological suggestions include pre- and peri-natal difficulties [35].

Neurobiological treatments

The main group of drugs used in patients with TS are the neuroleptics including haloperidol. All of these work by blocking dopamine receptors in the brain, although newer drugs have slightly different profiles and side effects. The response to drugs is variable and tics are rarely fully extinguished. Double-blind placebocontrolled designs have found tic frequencies reduced by approximately 50% using haloperidol in children and adults [5]. For further details on neurobiological treatments see [2,3,36].

Recent modern neurobiological treatments have been shown to be successful in treating patients with TS. For example, Mantovani was the first to report on the successful use of transcranial magnetic stimulation in TS [37].

Nagai *et al.* examined how changes in sympathetic arousal, induced using galvanic skin response biofeedback, impacted on tic frequency in 15 individuals with TS [38]. Results showed that tics (recorded via video) were lower during relaxation biofeedback when compared with arousal biofeedback, with tic frequency positively correlating with sympathetic arousal during the arousal session.

Hariz and Robertson reviewed the literature on deep brain stimulation in TS, which is only given for treatment resistant and severe TS in adults whose quality of life has been severely reduced and ideally in research settings [39]. They reported that there have been only two small deep-brain trials encompassing 50–55 cases, undertaken in 19 centers, and nine different brain targets have been used so far. Their conclusions were that the brain areas targeted are unclear and ideally this type of treatment should be restricted to research by experts only at present.

Psychoanalytic models

Although the first descriptions of tics related to a neurological model for TS, some early literature describe tics as psychological disorders and were considered to be sexual in nature. Mahler and Luke suggested tics as "erotic and aggressive instinctual impulses... which are continually escaping through pathological discharge", the treatment warranted by this model was aimed at giving the patient insight (allowing him/her) to realize that these impulses were the cause of his tics [40].

Psycholoanalytic treatments

The earliest psychoanalysts who treated individuals with TS included Ferenczi who felt that tics were equivalents of 'onanism' and both sexual and narcissistic in nature [41], Abraham who felt that tics were 'anal' rather than erotic in origin [42] and Klein whose tics were rooted in masturbation fantasies [43]. Mahler and her colleagues [40] perpetuated the psychoanalytic way of thinking about treatments of TS as did others (Kanner, Boncour and Reich), highlighting repressed sexual desires and a moral conflict between the ego and super-ego [44]; in the 1950s when Heuscher drew attention to both intrapsychic and environmental events and suggested that tics represented the struggle between the two [45].

Despite the use of psychoanalysis for tics, there have ever only been case reports and series published in the literature (Table 1).

Despite the lack of evidence-based studies on psychoanalysis or psychotherapy, these treatments may be useful and can increase capacity for self-reflection, improve better tolerance and frustration, bring about self-understanding and raise self-esteem [46]. Therefore, these forms of treatment are indicated for an individual's mental well-being, but are not indicated for tics *per se* (Table 1).

Following on from psychoanalysis, there were a number of attempts at other forms of psychological therapies such as hypnotherapy [47], awareness training [48] and massed negative practice [49] but only case reports or series were ever published. Table 1 contains a summary of some psychological therapies published in the form of case reports and series.

For an early general review on psychobehavioral therapies, please see references [44,46].

Behavioral models

A behavioral model for tics was suggested initially by Azrin and Nunn [50] and later modified by Turpin [51]. Azrin and Nunn suggested that tics develop following physical injury or psychological trauma and fulfill a function since they relieve muscle tension resulting from the injury or trauma [50]. They suggested that under normal circumstances, the tic would be inhibited by social awareness but sometimes it is not recognized and the movement is performed so often as to become a strongly established habit [50]. The repeated execution of the tic strengthens the specific muscles required for the tic and at the same time, the opposing muscles become relatively unused. Tics are then negatively reinforced by the tension reduction that follows the occurrence of the tic behavior or by other external factors such as social attention.

This model was modified when it was later discovered that many individuals are aware that a tic is about to occur. In 1980, Bliss described unpleasant sensory experiences that precede tics, the PSP mentioned earlier, that are relieved once the tic has occurred [21]. Turpin suggests that it is the sequence of the PSP, followed by tics that relieve the sensations, which may account for tic maintenance [51]. The author suggests that tics are conditioned responses that will be performed whenever a new sensation appears. In addition, he suggests that different kinds of stimuli may be associated with the sensory stimuli.

Behavioral treatments Habit reversal

Azrin and Nunn developed a package of interventions called habit reversal (HR) and it is currently referred to as habit reversal training (HRT) [50]. HR involves identifying early signs that a tic is imminent and then individuals are taught to produce an incompatible physical response contingent upon the urge to perform a tic. The tic-opposing muscles are contracted until the urge to tic has passed. Examples of an incompatible physical response include: for an eye-blinking tic, systematic voluntary, soft blinking consciously maintained at a rate of one

Table 1. A summary of published case reports and series of psychological and behavioral therapies for tic disorders and	
Tourette syndrome.	

Author (year)	Modality & type of treatment	Publications	Comments & conclusions	Ref.
Ferenczi (1921) Abraham (1921) Klein (1925)	Psychoanalysis	Psychoanalysis	Used successfully in the early part of the 20th century. The authors feel it is contraindicated unless the therapist is an expert in TS and psychoanalysis	[41] [42] [43]
Mahler and Luke (1946)		Psychoanalysis	Only slight/no improvement in 40% of ten patients	[40]
MacKay and Heimlich (1972)	Psychotherapy	Psychotherapy	This psychotherapy included 'paraverbal therapy', which was successful in one patient	[78]
Tophoff (1973)		SP	Not useful in a bright 13-year-old boy (IQ 120)	[79]
Lahey and McNees (1973)	Time out procedure	Time out at school	Elementary school teacher controlled the coprolalia and motor tics of a 10-year-old boy by giving him 'time out'	[80]
Canavan and Powell (1981)		Time out	No response in 24-year-old woman with TS	[81]
Doleys and Kurtz (1974)	Token economy	Token economy	Case report	[82]
Culbertson (1989)		Four-step hypnotherapy	Single case report treated for nine sessions – helped and well at 6-month follow-up	[83]
Lambert and Christie (1998)	Social skills group	Social skills group	Successful use of social skills in a group setting	[84]
Mansdorf (1986)	Assertiveness training	Assertiveness training	Reported that symptoms were in remission even at 1-year follow-up	[85]
Tansey (1986)	Biofeedback	EEG sensori- motor rhythm biofeedback training	Simple and complex tics were eliminated via EEG biofeedback. However, no long-term follow-up and studies not replicated.	[86]
Thomas <i>et al.</i> (1971)	SM	SM and reciprocal inhibition	Serendipitously found SM to be effective in a young male with TS. This procedure was based on the self-awareness of tics	[87]
Hutzell <i>et al.</i> (1974)		SM	Five-step successful SM in 11-year-old boy with TS (with loud barking)	[88]
Billings (1978)		SM	Successful treatment of 17-year-old treatment-naive female, with reduction of tics at 6-month follow-up	[48]
Ollendick (1981)		SM	Two children responded, one to SM alone and one with other treatments	[89]
Feldman and Werry (1966)	MP	MP	Unsuccessful case report of MP in a person with tics	[90]
Nicassio <i>et al.</i> (1972)		NP	Two patients treated with MP/NP. The first (22-year-old male) with a single neck tic responded well; the second (33-year-old male) with multiple tics failed to respond: he had failed to respond to numerous treatment modalities (medication, ECT, psycho-drama, group therapy and psychoanalysis)	[91]
Tophoff (1973)		MP, relaxation and assertion training	Package of treatments in a 13-year-old patient. Social contingencies were also taken into account. He recovered after 14 sessions	[79]
Knepler and Sewall (1974)		NP	NP paired with smelling salts in the treatment of a tic - case report	[92]
Storms (1985)		Massed NP	Reported succesful treatment in two cases of TS	[49]
Azrin <i>et al.</i> (1980)		NP (NP vs habit reversal)	Only randomized study including 22 patients with tics recruited through media advert. Ten out of 22 had HRT and 12 out of 22 had NP. HRT = 97% reduction at 18 months. NP only successful in approximately 30%. Overall insufficient evidence for MP	[93]
BFA: Brief functional an ECT: Electroconvulsive SM: Self-monitoring; SP Data taken from [44,62	alysis; CCR: Cue controlle therapy; ERP: Exposure a : Supportive psychothera].	d relaxation; CR: Competing nd response prevention; HR apy; TS: Tourette syndrome.	response; DR: Differential reinforcement; DRO: Differential Reinforcement Of zero-rate Behavior; Habit reversal; HRT: Habit reversal training; MP: Massed practice; NP: Negative practice; 	

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Table 1. A summary of published case reports and series of psychological and behavioral therapies for tic disorders and Tourette syndrome (cont.).

Author (year)	Modality & type of treatment	Publications	Comments & conclusions	Ref.
Rosen and Wesner (1973)	Relaxation training	Relaxation	Case report	[94]
Tophoff (1973)		Relaxation	Useful only as part of package	[79]
Canavan and Powell (1981)		Relaxation training	Case report. No response to relaxation, MP or oral feedback	[81]
Turpin and Powell (1984)		CCR	Reports of three single case studies. MP produced no reduction of tics in any case. CCR reduced tics in only one person	[95]
Zarkowska <i>et al.</i> (1989)		CCR technique	Case report of technique used on girl with mental handicap (learning disability) and not useful outside sessions	[96]
Bergin <i>et al.</i> (1998)		Relaxation therapy	A total of 23 patients included in a controlled study (Table 2)	[63]
Peterson and Azrin (1992)		Relaxation	Review of seven studies not mentioned here – tics reduced only temporarily	[97]
Evers and Van de Wetering (1994)	Behavior therapies (NOS)	Tension-reduction technique	Case reports of two patients who responded successfully to this treatment	[98]
Watson and Sterling (1998)		BFA	BFA useful in a single case study (patient coughed whilst eating).	[99]
Carr and Bailey (1996)		Brief behavior therapy	Tics were decreased by almost 70% and this was maintained at 1-month follow-up	[100]
Wagaman <i>et al.</i> (1995)		DR	Useful in a 9-year-old boy and reduced coughing and throat clearing 50 weeks post-treatment	[101]
Woods <i>et al.</i> (2000)	ERP	ERP	ERP useful in a 16-year-old boy with touching (thought to be a compulsion). ERP also useful in anxiety. Patient in residential care. Touching reduced 5 months after follow-up	[102]
Wetterneck and Woods (2006)		ERP	Useful in three repetitive behaviors in an 11-year-old boy	[103]
Finney <i>et al.</i> (1983)	CR and HRT	HRT	Two adolescent boys treated with HR in addition to a simple breathing and muscle relaxation exercises. Videotapes (in the homes) were used to show a 50–98% reduction in tics, which was supported by others' judgements such as teachers. Response maintained at 5 and 12 months	[104]
Miltenberger et al. (1980)		Contingent vs	Components of HRT. Results showed that noncontingent CR was ineffective, but contingent CR was effective. HRT also good. Small study	[105]
Sharenow <i>et al.</i> (1989)		Dissimilar competing response	Three patients with inconclusive results	[106]
Tolchard (1995)		HRT	HRT successful in a single case study	[107]
Woods <i>et al.</i> (1996)		Abbreviated HRT	The four children studied had variable responses to parts of HRT	[76]
Woods <i>et al.</i> (2003)		HRT	Four out of five children responded. HRT significantly better than other therapies for tics	[108]
Himle <i>et al.</i> (2008)	Differential	DRO	This study reported that three out of four children had lower tic frequency	[109]
	reinforcement		when tokens were delivered in a DRO exercise, namely when tokens were	
			delivered contingent on the absence of tics (than when tokens were	
			delivered noncontingently)	
BFA: Brief functional an	alysis; CCR: Cue controlle	d relaxation; CR: Competing	response; DR: Differential reinforcement; DRO: Differential Reinforcement Of zero-rate Behavior;	

ECT: Electroconvulsive therapy; ERP: Exposure and response prevention; HR: Habit reversal; HRT: Habit reversal training; MP: Massed practice; NP: Negative practice; SM: Self-monitoring; SP: Supportive psychotherapy; TS: Tourette syndrome. Data taken from [44,62].

> blink per 3–5 s; for arm movements, pushing hand down on thigh [52]. Support from family members for tic-free intervals and adherence to protocols is also part of the overall package.

The mechanism of action of these types of therapy were first documented by Azrin and Nunn [50] when they suggested that treatment effects were due to strengthening of muscles, that are incompatible with tic expression. They treated 12 patients with HR who had a variety of 'nervous habits' (including tics [n = 3; ages 14, 14 and 64 years]). They were trained with HRT (awareness training, competing response practice, habit control motivation and generalization training) and the tics disappeared in two and in the other reduced from 8000 to 12 per day.

Azrin and Peterson then carried out the first randomized controlled trial looking at HRT versus a waiting list control group in ten subjects with TS [53]. The majority of subjects were under 16 years old and two were younger than 10 years old. The group contained most subjects under 16 and two under ten. Tics decreased on average by 93% in the HR group. Weaknesses in the sample selection included the fact that some children were on medication at the time of the study, comorbid conditions had not been excluded, and there was a wide variation in the number of sessions offered (13-30 sessions). The outcome measures were taken at 12 months from the start of treatment and, therefore, some individuals were still having treatment 1 month prior to scoring, making it difficult to compare outcomes meaningfully. There was also no long-term follow-up.

Wilhelm et al. carried out a randomized controlled trial reporting that HRT was more effective than supportive psychotherapy in TS [54]. Although outcome scores were better than pretreatment scores at 10-month follow-up, there was no significant difference in scores between groups. Authors say this was owing to the fact that tics increased slightly in the HRT group and decreased slightly in the waiting list group. This paper was published as a brief report rather than a detailed paper so it is suspected that some details were not included. The age range is not given hence it is unclear how many, if any children were used in the study. The assessors were not blind to treatment groups, meaning there may have been some bias in scoring, but the clinical global impression scores [55], which were subject-rated, reflected the assessors' findings. The authors raise questions regarded the long-term effects and suggest the need to include relapse prevention and booster sessions.

Recently Piacentini *et al.* published results of a two-phase multicenter, randomized controlled trial for HRT in 126 children and adolescents [56]. Phase 1 involved looking at treatment response between HRT and supportive therapy. The HRT treatment follows a structured manualized protocol as set out in Woods *et al.* [57]. Phase 2 involved a 6-month observation period of both groups. Three different sites were used. The aim of the study was to evaluate whether the manualized HRT approach was superior to supportive therapy and education for reducing tics. Participants were aged between 9 and 17 years with a diagnosis of Tourette or chronic tic disorder and 78% were boys. Comorbid conditions such as attention deficit hyperactivity disorder were included so long as the condition was stable and did not need a change in current treatment. Children on medication were also included so long as the medication did not need changing. The HRT also included a form of relaxation training and a 'functional intervention' to deal with stressful situations that may contribute to the worsening of tics. The article gave an example of parents being taught to specifically praise their children for practicing HRT on returning from school when tics are particularly more intense. The supportive therapy involved education regarding tics but therapists did not discuss strategies to manage tics. Both interventions were carried out over 10 weeks delivered in eight separate sessions. Therapists also received weekly supervision across sites to maintain integrity of interventions. Intervention integrity was also maintained by a sample of videotaped sessions being checked.

Results showed that after 10 weeks, HRT caused a significantly lower Yale Global Tic Severity Score compared with supportive therapy. HRT was associated with a 7.6-point decrease in YGTSS compared with 3.5-point decrease in the supportive therapy group. The authors state this four point difference in scores is comparable with that of medication and placebo in some medication trials. The psychosocial outcome as measured by the clinical global impressions score was also significantly reduced for the HRT group.

Exposure response prevention

Exposure-based interventions (ERP) are based on the conceptualization of tics as voluntary intentional movements that are performed in order to decrease unpleasant sensory urges experienced in muscles. ERP entails exposure to the sensations and urges that precede tics, and response prevention of the tics. The patient habituates to the premonitory experiences, thus resulting in tic reduction [58].

Verdellen *et al.* compared ERP and HRT. The authors studied 43 subjects with TS who were randomized to treatment groups [59]. Some individuals were taking medications and these individuals were also randomized separately so both groups had equal numbers of medicated subjects.

Assessors were blind to the treatment applied. Results showed that both HRT and ERP showed significant improvements on all tic outcome measures after 3 months. No differences between the two interventions were reported.

Although the study was randomized, it is not clear if age was a factor in the randomization. This is important given the mixed age cohort. It is difficult to comment on long-term benefits as follow-up was at 3 months.

Relaxation training/therapy

Tics often increase with stress and anxiety [60,61] and, therefore, any efforts to reduce stress should lead to a reduction in tic symptomatology. Relaxation training (RT) involves deep breathing exercises and guided imagery [51] and the main aims with this type of treatment are to reduce stress and alleviate anxiety [62].

Various case reports (Table 1) have shown moderate reductions in tic frequency but in the majority of well-controlled designs relaxation has not been effective when used alone as a single treatment strategy.

One of the two controlled trials was that of Bergin et al. who carried out a randomized trial comparing RT versus minimal therapy (control group) [63]. Minimal therapy was defined as 'awareness and quiet time'. The authors examined 23 children from a pediatric TS clinic, of whom 16 completed the 3-month study. Children were randomized according to tic severity and presence of comorbid attention deficit hyperactivity disorder. Although there was an initial improvement in the relaxation group (n = 7), there was no difference between groups at the 3-month follow-up. The authors concluded RT has a limited role in treatment of tics. Strengths of the study include assessors being blind to treatment groups and that several objective outcome measures were used.

Despite these results, and because some people with TS have an increase of tics with a wide variety of environmental stressors [60,61] relaxation training is often used as part of a comprehensive treatment package [62].

Massed practice

Massed practice (MP) has been used for many years as a treatment of tics [64,65], the basic thesis being over-rehearsal of the target tic and, therefore, the patient voluntarily reproduces and repeats tics several times a minute. This overrepetition is supposed to produce a build-up of inhibition/fatigue and subsequently results in tic reduction [62]. As can be seen from Table 2 there have been numerous case reports using MP (with mixed results), but only one randomized trial. In this study, Azrin and Peterson, who have been acknowledged as the main instigators and proponents of the technique, advertised in the media and included 22 subjects with TD in a trial of HR (n = 10) versus MP (n = 12) [53]. HR decreased tics by 99% at 4-6 months and 97% at 18-month follow-up. Rating was carried out by observers studying video evidence of tics. HR was more effective than MP, which only reduced tics by 33% at 4 weeks.

A cognitive model for tic disorders

A cognitive model for tic disorders has been described by O'Connor [22]. The basis of this is that tension increases the likelihood of tics and that thought and emotions have an effect on tension. The model proposes that tics always occur in certain situations, which represent to the person a sense of frustration. Cognitive appraisal of the situation is distorted, which contributes to the person feeling tense [22]. O'Connor suggests that individuals with TS/TD have core perfectionist beliefs regarding self-image, personal standards and personal organization [22]. This, then, creates unrealistic expectations of performance, which lead to a counterproductive style of action (e.g., attempting too much). Subsequently, frustration at not performing as desired, produces heightened tension, part of which involves over preparing muscles for the anticipated task (associated with the frustration). This tension can be reduced short term by repetitive tense-release cycles, which then provokes the tic [22]. The model also suggests a complex interaction with hypersensitivity to muscle sensations, which in turn causes increased restlessness, which adds to increased tension.

As a result of the model, it is suggested that cognitive therapy (CT) might change and confront expectations involved in the way the individual prepares for a situation, and so modify how the person acts in the situation [22]. Cognitive strategies would challenge overactive style of behavior, effortful preparation in anticipation of high-risk situation hyper-attentiveness to self and excessive self-consciousness and concern over self-image.

Table 2. Publi	shed controlled tr	rials of	behavioral t	reatments i	for tic disor	ders.				
Author (year)	Design	c	Age range (years)	Diagnosis	Group	Outcome measures	Treatment information	Follow-up	Outcome	Ref.
Azrin <i>et al.</i> (1980)	Controlled	22	11–62	TS and TD	HR vs NP	Self and family reports	1–2 sessions HR 1 session NP	18 months	HR significantly better than NP	[93]
Azrin and Peterson (1990)	Randomized	10	6–36	TS	HR vs WL	Observer rated videotape tic frequency clinic and home	Varied: an average of 20 sessions⁺	12 months from start of treatment	All tics in HR decreased by 93% at home and clinic	[53]
O'Connor et al. (1997)	Randomized	14	23-49	CTD	CT vs HR	Self-report of tic intensity and frequency Personal reaction to tics EMG under imaginal risk condition	Weekly sessions for 10 weeks	3-month and 2-year telephone contact	Both groups showed decreased tic frequency by 22 and 21% at 3 months, respectively No significant difference between groups, 50% from both groups maintained improvement at 2 years	[66]
Bergin <i>et al.</i> (1998)	Randomized	23	7–18 Mean: 11.8	TS	RT vs ST	YGTSS TSSS GVS Assessors blind	6 weekly sessions	3 months from start of treatment	Post-treatment improvement No difference between groups at follow-up	[63]
O'Connor et al. (2001)	Not randomized	128	CTD mean: 39.1 HD mean: 37.1 Children excluded	HD HD	HR vs WL HR vs WL	Degree of control Self-rated tic/habit frequency and intensity Video rated blind	4 months of weekly sessions	2-month and 2-year telephone contact	CTD and HD in treatment group showed improvement in tic frequency and intensity 65% had 75–100% control No difference between CTD and HD Of those seen at 2 years, 77% maintained improvement 52% obtained 7–100% control	[67]
Wilhelm <i>et al.</i> (2003)	Randomized	32	Mean: 36.2 for HR and 33.2 for SP Range not provided	TS	HR vs ST	YGTSS observer rated CGI self-rated	8 weekly sessions then 2 per month A total of 14 sessions	5 and 10 months	Tic scores decreased in HR compared with control group but significance not shown at 10-month follow-up CGI scores were significantly better for HR group at 5 months	[54]
⁺ See [13–30] . CBT: Cognitive-ber HR: Habit reversal; J WL: Waiting list; YG	iavioral therapy; CGI: Clir NP: Negative practice; R1 .TSS: Yale Global Tic Seve	nical glok T: Relaxat erity Scale	al impression; CT ion therapy; SD: 5	: Cognitive then Standard deviati	apy; CTD: Chroi on; SP: Support	ric tic disorder; EMG: Electromyogra, ive psychotherapy; ST: Supportive th	phy; ERP: Exposure res, nerapy; TD: Tic disordei	ponse prevention; GV: rs; TS: Tourette syndro	S: Goetz videotaped scale; HD: Habit disor me; TSSS: Tourette Syndrome Severity Scal	der; le;

Table 2. Publis	shed controlled	trials of	f behavioral ti	reatments f	or tic disor	ders (cont.).				
Author (year)	Design	٢	Age range (years)	Diagnosis	Group	Outcome measures	Treatment information	Follow-up	Outcome	Ref.
Deckersbach et al. (2006)	Randomized	30	Mean: 36.6	TS	HR vs ST	YGTSS observer rated CGI self-rated	8 weekly sessions	6 month	Tic scores decreased in HR compared with control group Both treatments improved on CGI	[011]
Verdellen et al. (2004)	Randomized	43	7–55 Mean: 20.6 SD: 12.1	TS	ERP vs HR	YGTSS videotaped sessions Tic frequency monitored via YGTSS observer rated CGI self-rated Videoed sessions Assessors blind	12 weekly ERP sessions 10 weekly HR sessions	3 months	YGTSS HR effect size 1.06 YGTSS ERP effect size 1.42 No significant difference between two groups	[59]
Piacentini <i>et al.</i> (2010)	Randomized	126	9–17 Mean: 11.7 SD: 2.3	TS	HR vs ST	YGTSS observer rated CGI self-rated	8 sessions weekly then 3 monthly booster sessions	3 and 6 months	Tics and CGI scores decreased significantly in HR group	[56]
+See [13–30]. CBT: Cognitive-beh HR: Habit reversal; ML: Waiting list; YG	avioral therapy; CGI: Cl NP: Negative practice; I TSS: Yale Global Tic Se [:]	llinical glol RT: Relaxa: verity Scal	bal impression; CT: tion therapy; SD: S ⁱ le.	: Cognitive thera tandard deviatic	apy; CTD: Chroi on; SP: Support	nic tic disorder, EMG: Electromyogre ive psychotherapy, ST: Supportive t.	aphy; ERP: Exposure res herapy; TD: Tic disorde	sponse prevention; GVS ·rs; TS: Tourette syndror	s: Goetz videotaped scale; HD: Habit dise me: TSSS: Tourette Syndrome Severity Sc	order; ale;

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Cognitive-behavioral therapy

There have been two controlled treatment studies in which the cognitive model has been used as an intervention in TS. O'Connor et al. examined patients with TD, allocated to either an HRT treatment group or a CT treatment group [66]. Follow-up was at 3 months after treatment and telephone contact 2-years post treatment. Muscle activity was recorded on the tic-affected muscle via electromyography (EMG) during the third session and post-treatment. The first five sessions included awareness training and monitoring of tics and muscle relaxation. In session five subjects were asked to identify a high-risk situation in which tics occurred. Sessions six to ten involved either an HRT package as in Azrin and Peterson [53] or CT aimed at modifying anticipations identified earlier. The CT themes were based from personal constructs collected in session five. Any distorted cognitions or maladaptive expectations were confronted and replaced with more realistic anticipations using Socratic dialog. The primary focus was on changing the way the person evaluated his/ her, actions in the high-risk situation. Assessors were blind to treatment groups. Self-report on frequency and degree of control over the tic was also measured. Both groups showed statistically significant improvement in tics at 3 months, with no difference between groups. EMG recording also decreased. At 2-year follow-up, three of the HRT group and three of the CT group had maintained improvement. Two of the CT group could not be contacted. The authors conclude that cognitive therapy is helpful.

The use of EMG as a physiological marker for change was a helpful and novel outcome measure and it was also helpful having outcome measures after 2 years; however, this was only telephone contact thus there may have been subjective bias. Only three of the HRT and three of the CT group maintained improvement at follow-up, questioning the long-term benefits of both treatments. It is also not clear whether the benefits were due solely to the cognitive components of the package or other factors such as awareness training and relaxation. Limitations to the study included small sample size and that the study used a nonclinic population, thus limiting the usefulness of the study as tics are likely to be less severe than in those referred to health services.

O'Connor *et al.* examined the efficacy of a cognitive-behavioral therapy (CBT) program based on HRT for the management of chronic

tic disorder (CTD) and habit disorder (HD) in adults [67]. In total, 47 subjects received a 4-month treatment program while 38 were on a waiting list. Participants were recruited via an advert in a local newspaper. Habits included bruxism (teeth grinding) and trichotillomania (hair pulling). Baseline measures include tic intensity and frequency and degree of control over tic. A situation grid identified potential high- and low-risk situations. Video ratings were taken 10 mins before and after rest and then during imagination of a high-risk scenario. Independent assessors who were blind to the treatment groups rated the videos.

The CBT package was similar to the HRT protocol mentioned in Azrin and Nunn with CT added [50]. Socratic dialog was used to confront unrealistic anticipations concerning reactions of others, own performance and appearance in high-risk situations. Results showed that CBT was more effective than the waiting list group, and was equally effective for tic and HDs. The sample studied was unlikely to have tics that were as severe as a clinic population as they were recruited via advertisements. Strengths of the study included comorbidity being excluded, and objective measures employed. However, video rating was only achieved in 63 subjects and there were also some missing diaries. The authors conclude that CT is a useful addition to HRT. A major weakness in the paper is that it does not give any details as to how the groups were allocated, and thus it is not clear whether there was any degree of randomization. This makes it difficult to interpret findings in the context of evidence-based research.

Although not directly comparable, one is tempted to learn from the Turner et al. pilot study of telephone-CBT employed in young people with OCD [68]. OCD, as has been previously mentioned, is highly comorbid with TS, and in the UK CBT is the recommended treatment for OCD in young people. Access to face-to-face CBT (for both TS and OCD) may be limited by a number of factors including the number of trained therapists, as well as both geographic and financial resources preventing access to specialized services in both the NHS (or equivalent) and private sector, internationally. Turner et al. embarked on a pilot study of telephone-based-CBT for ten young people (aged 13-17 years) with OCD attending a specialist clinic, in which the young people and their parents received up to 16 sessions of telephone-based-CBT [68]. The measures of OCD symptoms were obtained using multiple informants and a repeated measures design. The assessments were conducted at pretreatment, post-treatment and also at 6- and 12-month follow-up. Results showed that OCD symptoms improved according to all informants and also family satisfaction with the treatment modality was high. This method may well be considered for adaptation for patients with TS.

Although there are now several randomized controlled studies looking at psychological treatment in TS, no studies have directly compared psychological intervention with medication. For a summary of all randomized controlled trials involving psychological therapies see Table 2.

Developmental issues in young people (prepubertal) with tics & TS

Management of both TS and tics is potentially more difficult in younger people as firstly they have not yet fully developed (and thus have developmental issues that have to be addressed), but also medications and their adverse side effects have not been fully understood and documented in many settings, including TS, with particular reference to long-term follow-up and side effects.

When examining the psychological therapies in children, there is also a need to consider the specific therapy component that is utilized, that is to say, behavioral interventions or cognitive interventions [69]. If therapies rely on cognitive interventions then consideration needs to be given to developmental issues. Durlak et al. [70] found that children aged 11-13 years derive significantly more benefit from CBT than younger children. Traditional Piagetian theories of cognitive development also suggest that it is only when children are in the formal operational phase of childhood that they are able to think in abstract terms and, therefore, anyone under 12 would have difficulty engaging in CBT. Flavell et al. reported that 8 year olds and adults could link thoughts to feelings but 5 year olds could not [71]. The work by O'Connor et al. emphasizes cognitions such as appraisals and thoughts around perfectionism, which, even if these cognitions were present, may be difficult to assess in young children [67]. Studies involving HRT and ERP do not involve complex cognitive processes, so one may be able to apply these therapies to children.

Holmbeck *et al.* make a number of suggestions for researchers when considering developmental issues in evidence-based practice [72]. He states that "prior to developing or evaluating a treatment, it is critical that an investigator conceptualize the disorder in question from a developmental perspective". With this recommendation in mind, the developmental issues in TD are important. One of the essential aspects in psychological therapies is awareness of tics and in particular the ability to recognize the PSP.

The individual learns to use the PSP for preventing the actual tic performance through a competing motor response or by shaping strategies to reduce intensity and intrusiveness of tics. The question thus arises, 'at what age are clients able to detect a PSP?' Leckman *et al.* [20] found 93% of adults with TS reported PSP while Kurlan *et al.* [73] found PSP reported in 74% of adults with TS. Leckman and coworkers remarked that young children under the age of 10 years with simple tics (e.g., eye blinking, quick head jerk) usually do not have PSP, or are totally unaware of these sensations [74].

Banaschewski et al. carried out a cross-sectional survey of 254 children and adolescents (age range 8-19 years) with TS to investigate the developmental aspects of PSP [75]. To test for developmental effects, the total group was stratified into three age groups: 8-10, 11-14, and 15-19 years. Of the 254 subjects, 37% reported PSP. This is much lower than reported in previous studies involving adult patients [20,73]. A look at the results of the three different age groups studied shows that the rates for awareness of PSP increased from childhood to adolescence. Statistically significant stepwise increases were found at two different age levels. The essential maturational step was observed between the group of 8-10 year olds and the 11-14 year olds. Banaschewski et al. conclude that the natural course of tic awareness increases with age [75].

Therefore, psychological therapies such as HR and ERP that rely on the presence and awareness of PSP would probably be more appropriate for the older adolescents than younger children.

Another issue in relation to therapy for children and adolescents with TD is whether the young person is affected by tics to the extent that they wish to do something about it. There has to be a degree of motivation for psychological therapies to work and from clinical experience, it is usually the parent who brings the child to the clinics and the child is not too concerned. When the child is older and more aware of social aspects and peer group pressure, the young person may wish to do something about it.

Discussion

Despite numerous case reports and case series purporting successful outcomes for psychological therapies, there are at present only ten studies that use controls as part of its design. There are methodological limitations that need to be considered when comparing these studies. These include small sample sizes, use of medication, clinic versus nonclinic cohorts and variations in outcome measures. It is also not clear what effect comorbid conditions have on the efficacy of intervention. Some studies examined pediatric populations [56,63], whilst others included a mixed sample of children and adults. When is the best time to measure outcome and for how long? Some studies report tic reduction by as much as 99% but clinical and academic experience has shown us that no intervention, whether pharmacological or psychological has ever been able to reduce tics by that amount for a significant period of time. Thus objective measures over a period of time would be a more appropriate measure of outcome effectiveness.

It appears that HRT is effective in TD/TS and this is more apparent in the recent randomized controlled trial by Piacentini *et al.* but there is a need for further trials [56]. Altogether, three studies showed success with HRT when compared with a waiting list group or a supportive therapy group. Two further studies also showed successful outcomes when HRT was used as a control group. However, there is some doubt about the long-term benefits of HRT and it may be that relapse prevention and booster sessions are needed.

The study by Piacentini *et al.* should be highlighted as it did include children who were on medication and had comorbid conditions that were stable thus suggesting applicability to the population in 'real life' clinics [56].

The operational components and treatment parameters of HRT also need to be explored in more detail as it is still not clear from the literature which components of HRT are necessary for symptom reduction [76]. Although HRT studies included some children in its sample, it is not clear whether it is a useful therapy for children due to the developmental issues outlined earlier. Both HRT and ERP use specific awareness of tics such as the PSP as the starting point for their intervention. Verdellen *et al.* achieved relative success using ERP in children and adults [59].

The relaxation study by Bergin *et al.* showed improvement at the end of treatment but this was not sustained long term [63]. Given that this technique is relatively easy for children to utilize, it may have a role with short-term agendas such as when a child has to perform on stage or public speaking.

Only two studies looked at the cognitive aspects of TD. Given that some tics worsen with stress and some worsen in specific situations, one might assume that altering cognitive thoughts may have an effect on tic symptomatology.

The cognitive model is interesting but the model needs to be tested appropriately. Much more work is needed before firm conclusions can be made about the effectiveness.

It is unclear whether the cognitive model could be applied to children given the developmental issues mentioned earlier. In order to generate a useful model, a thorough assessment is needed in this specific client group. This would include identifying cognitive processes such as negative automatic thoughts, cognitive distortions, core beliefs, and secondary appraisals [77]. At present, no one has examined these processes and other situational factors in children and adolescents with TD/TS. It would also be interesting to look at potential family and cultural influences such as parental views and attitudes around tics as well as individual beliefs regarding causes of tics.

In general, the literature does not distinguish between isolated tics that develop in adulthood, say for example a piano player who develops a tic in his hands, and the multiple tics that develop usually in childhood and are part of a neurodevelopmental disorder such as TS. Putting both conditions together in the spectrum of TD is useful but it does not imply that they have the same etiology. It would be essential for both behavioral and cognitive models to address this issue.

The treatment literature also needs better clarification of the actual intended outcome of treatment. Is the aim to reduce tics for a short period of time or to completely cure the person of tics? Since tics wax and wane, it makes it difficult to determine whether changes in the tics are the result of treatment effects or part of the natural course of the disorder. Only two studies had follow-up greater than 1 year [66,67], although this was done by telephone contact introducing subject bias. The possibility of spontaneous remissions and the erratic course of tic symptoms would suggest the need for repeated post-treatment assessments. If psychological therapies were found to be effective, then services of course should be providing these treatment options to children and adolescents. However, at this stage there is no firm conclusion on long-term benefits of any of the therapies.

Recommendations for future studies concerning children and adolescents with TD include:

- Studies looking closely at the specific degree to which operant variables affect tics in children and adolescents
- More research identifying related cognitive factors
- Further randomized controlled trials looking at HR and/or ERP in children and adolescents with TD who experience PSP
- Longer-term follow-up

Conclusion & future perspective

In summary, psychotherapy and analysis are not useful for tics but may be useful for other issues such as depression or poor self-esteem: however, we recommend that the latter therapies be employed by those clinicians who specialize in TS as well as psychotherapy/analysis. The plethora of other therapies shown in Table 1 are probably not useful in TS and have only historical interest. Although MP has been documented many times, there is insufficient evidence to date for this method being used as a management for tics in patients with TS. There is some evidence that RT can be used as part of a 'package'. SM is also not indicated for monotherapy. There is some evidence that ERP may be useful for TD/TS. However, there is some uncertainty about long-term effectiveness of these methods. CBT/CT may be useful, but relaxation therapy/training is only really useful as part of a comprehensive treatment package.

From the smaller trials and also ten controlled trials, it does seem that HRT is a good and effective treatment for the tics of TS and would be given an 'A' if the same criteria were applied as to the drug trials (this is the criteria given to many drug trials, see [36]). Therefore, the psychological treatment that can be considered to have positive benefit would be HRT.

Despite the new emerging evidence suggesting some psychological therapies may be beneficial in reducing tics, a practical difficulty is that in some countries (e.g., the UK), there are very few trained individuals who can undertake these forms of treatment, which do require a significant time and resource commitment. With the momentum of positive reported studies gathering pace, we would encourage interested clinicians, such as psychologists to explore HRT in more depth and seize the opportunity to train in this area and carry out

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trials. The next 5–10 years will be important if this form of intervention is going to take off.

In general, apart from HRT, more controlled trials are needed before firm conclusions can be drawn about the effectiveness of the behavioral therapies.

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