Invited essay

A cognitive-behavioral/psychophysiological model of tic disorders

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Abstract

This article discusses current cognitive behavioral, as well as neurophysiological, accounts of the development and maintenance of tic behavior in chronic (simple or complex) tic disorders. A cognitive psychophysiological model is further elaborated, highlighting the reciprocal interplay of background cognitive and physiological factors preceding tic onset. According to the model, cognitive factors such as perfectionist concerns and heightened sensory awareness and self-attention, as well as physiological factors such as a high level of motor activation and accompanying elevated muscle tension, play a role in tic habits. Negative appraisals of tics and counter-productive coping strategies developed by clients as a means to suppress or to disguise the tic behavior may also locally reinforce tic onset. Neurochemical factors are viewed largely as concomitants of behavioral adaptations or compensations to the tic problem rather than as independent markers or precursors of tic onset.

Clinically, the model emphasizes the role of cognitive-behavioral factors in tic onset, and suggests that tic management is best accomplished through cognitive behavioral interventions designed to prevent build up of both tension and pre-monitory urge in tic-affected muscles, rather than reverse the tic at the onset of the premonitory urge. The clinical validity of parts of the model is supported by recent experimental, psychometric and clinical studies. Other parts of the model remain speculative but at least yield testable predictions. A strength of the model is its ability to account for findings over diverse psychological and biological domains. © 2002 Elsevier Science Ltd. All rights reserved.

1. Problem

Tics are defined as repetitive non-voluntary contractions of functionally related groups of skeletal muscles in one or more parts of the body. Tics occur over all cultures, and have been reported...
anecdotally since classical times. According to Suetonius (119, 2000, Pg 177), the Roman historian and royal gossip, the Emperor Claudius had head and phonic tics. The first clinical descriptions however were provided in the 19th Century by Itard (1825) who reported a case of tics, barking and obscenities, and later Gilles de la Tourette (1885) who gave a detailed description of eight additional cases. Currently, the Diagnostic Statistical Manual of the American Psychiatric Association (DSM-APA, 1987) distinguishes transitory tics from chronic tic disorders (TD) and Gilles de la Tourette’s syndrome (TS). TS is recognized in the DSM-III-R & IV as a separate diagnostic category with multiple tics including vocal (phonic) tics occurring several times per day, and although clinician consensus tends to view TD as a milder form of TS, diagnosis of TS is categorical not dimensional. Tics may be simple or complex. Simple tics include blinking, cheek twitches, head or knee jerks and shoulder shrugs. Tics are mainly confined to the upper body and the most common occur in the eye, head, shoulders and face. Tics can also be vocal and include coughs, tongue clacking, sniffing, throat clearing, hiccing, barking and growling. Some recurrent involuntary somatic sensations are classified as sensory tics. These are identified as heavy, light, warm or tingling premonitory sensations, often muscle focused and leading to muscle tension (Lohr & Wisniewski, 1987; Shapiro & Shapiro, 1986; Shapiro, Shapiro, Young, & Feinberg, 1988) but the term “premonitory sensation” is now preferred over sensory tic (Cath, Spinhoven, Hoogduin, Landman, van Woerkom, van de Wetering et al., 2001).

Tics are classified as complex if there is a contraction in more than one group of muscles (Comings & Comings, 1984). Complex tics may involve sequences of movements, and may take the form of bizarre mannerisms involving limbs, head or extremities. Some complex tics can look like maladaptive normal responses. They may exaggerate a normal action, or include a redundant aspect in a normal action, for example by involving an irrelevant body part (e.g. exaggerated blinking, including cheek muscles). Complex tics may take the form of self-inflicted repetitive injurious actions such as head slapping, face scratching, tense-release hand gripping cycles, or dystonic posture. Complex vocal or, more precisely, phonic tics (Jankovic, 1997) take the form of repeated sounds, words or phrases or swear words, and in rare cases, swearing (coprolalia). Normal actions and words of the person may also be repeated, and copying others can itself evolve into a complex repetitive movement, either by motor mimicry (echopraxia) or by repeating others’ words, phrases or sounds (echolalia). Complex tics can resemble habit disorders (HD) such as trichotillomania, bruxism, scabioromania, onychophagia, which are, however, classified amongst the impulse control disorders. There is a covariation between tics and HD and amongst different types of HD (Woods, Miltenberger, & Flach, 1996).

Technically speaking, complex tics are distinguishable from stereotypes and compulsive rituals, routines and habits, since tics are neither completely conscious purposeful rituals, nor totally nonsensical repetitions. In fact, the term behavioral stereotypy is usually applied to abnormal repetitive actions associated with organic loss and mental deficiency. In practice, however, it is sometimes difficult to distinguish tics, rituals and repetitive movements. The relationship between these three is puzzling since some movements have impulsive as well as compulsive elements. Shapiro and Shapiro (1986) referred to “impulsive compulsions” to highlight the confusion, and Rasmussen and Eisen (1992) and other authors have equally underlined the importance of understanding the relationship between impulsion and compulsion for clarifying diagnosis. The diagram in Fig. 1 illustrates the distinction between habits, tics, and routines, in terms of two crucial dimensions of awareness and volitional control. In normal automated routines, there may be little
Fig. 1. Locates four types of actions: controlled responses, automatic routines, compulsive rituals and impulsive reflexes, in accordance with two orthogonal cognitive dimensions of voluntary choice and awareness.

awareness but there is overall volitional control. In rituals there may be awareness but no control. In reflexes and tics, there may be neither awareness nor control.

1.1. Secondary distress

Tics are rarely life-threatening except in rare cases where they may provoke auto-mutilation. Psychosocial distress, however, in this group can be considerable and can involve secondary phobias, depressions and social anxieties and worries over self-image, very low self-esteem and relationship problems. In our estimation of the interference of tic and habit disorders in daily activities, we found problems ranging from unemployment, marital conflict, interpersonal difficulties, employer relations, travel restrictions, problems attending social or public functions, performance worries (e.g. about driving, speaking, teaching, dancing, sport), all of which were perceived (by the affected person) to be a result of the tic habit (O’Connor, Brault, Loiselle, Robillard, Borgeat, & Stip, 2001). Such problems multiply for those with multiple tics, as has been documented by the Tourette association (see Green leaflet series, Canadian Tourette Association). People with tics often experience low self-esteem and are (or become) hyperattentive to the judge-
ment of others with consequent low self-satisfaction (Thibert, Day, & Sandor, 1995). Christenson, Faber, Zwann, Raymond, Specker, Ekern et al. (1994) also noted that in other impulsive disorders (e.g., compulsive buying), the person’s self-esteem seems to depend unduly on the response to others. Watson and Sterling (1998) reported the functional analysis of a case of a phonic tic, where the need for social attention was a precipitating factor. In TD, ironically, the very anticipation of experiencing a negative self-evaluation can provoke the tic (see later section).

1.2. Prevalence

The incidence of TS in adults is about 0.1–1%. The lifetime prevalence of TD is not known but estimates vary between 5 and 10% of the population. In a recent study of the Quebec population, O’Connor, Loyer, Lesage, and Robillard (submitted) found a self-report rate of 8% lifetime prevalence. Other recent estimates have placed the prevalence of TS at 1% and TD at 10% of the population (Robertson & Stern, 2000; Robertson, 2001). There are however problems with clinical estimates of prevalence. Fallon and Schwab-Stone (1992) pinpointed several methodological shortcomings in evaluating comorbidity studies of tic disorders, in particular sample selection bias (e.g., the clinician’s illusion springing from the use of clinical, not community, samples). Identifying cases through self-report can also be problematic, especially if people are misinformed about TS criteria. The “TICS” project underway, directed by D’ Roger Freeman in Vancouver, aims to standardize recording of all reported cases of TS worldwide, together with co-morbidity, to improve estimates of prevalence.

2. Assessment

2.1. Diagnostic instruments

There is no standard semi-structured interview (equivalent of the Anxiety Disorders Schedule [ADIS]) but individual researchers have developed their own interview protocols (e.g. Leiden protocol [Cath, Spinhowen, van Woerkom, van de Wetering, Hoogduin, Landman, et al., 2001; Robertson [2001]). The standard measure for clinical assessment of tic disorder is the Yale Global Tic Severity Scale (Leckman, Riddle, Hardin, Ort, Swartz et al., 1989). This scale records frequency, intensity, complexity and anatomical site of motor, sensory, phonic tics, and also interference in other areas of life. There are also other clinician rated scales such as the Tourette’s Syndrome Global Scale which scores degree of tic severity and assesses disruption due to tic multidimensionally (Harcherik, Leckman, Detlor, & Cohen, 1984). There have been attempts to adapt the Yale–Brown Obsessive Compulsive Scale (Y-BOCS) for use with tics and habits on the assumption that all form part of the same OC spectrum (Stanley, Prather, Wagner, Davis, & Swann, 1993), but the Y-BOCS is generally likely to underscore the presence of tics and habits.

2.2. Diagnostic boundaries

Estimates of comorbidity of TS with attention deficit hyperactivity disorder (ADHD) in children vary between 20–90% depending on diagnostic criteria (Robertson & Eapen, 1992) as compared
to 2–11% in non-TS populations (Costello & Angold, 1988). Both Comings and Comings (1984) and Shapiro and Shapiro (1986) felt ADHD was an integral part of TS. In adults with TD, as noted later, there also seems a particular style of action similar to hyperactivity, but with a clear cognitive component.

However, the two key diagnostic boundaries for TD–TS in adults are obsessive compulsive disorder (OCD) and HD. Some researchers consider all these disorders part of the same OCD spectrum. Hollander (1993) proposed that what unites the OC spectrum is an inability to inhibit or delay repetitive behaviors. There is some experimental evidence supporting the idea of a dimension between TS/TD and HD (O’Connor, Lavoie, Robert, Dubord, Stip, & Borgeat, 2001). Others argue for a distinct TS spectrum (Cath et al., 2001). On the face of it, there are phenomenological similarities between OCD, HD and TD. There is also a significant co-morbidity between TD, TS and OCD. The co-morbidity of a tic disorder with OCD in adults varies across studies from 25–63% and about 17% of adult OCD have tics (Holzer et al., 1994).

Cath and co-workers (Cath, Spinhoven, van Woerkom et al., 2001; Cath, Spinhoven, Hoogduin et al., 2001) have carried out a series of studies aimed to distinguish TS/TD from OC symptoms. A factor analysis of TS/TD, OCD±TD, and controls of OCD symptoms revealed distinct compulsive and impulsive factors. The OCD scored higher on the compulsive factor, and the TS/TD scored higher on the impulsive factor. When OCD-TD were compared to OCD±TD, the former scored higher on obsessionality. TS/TD patients reported more echophenomena, trichotillomania, touching, symmetry behavior, self-injurious behaviors, but less checking, repeating and rumination. Self-injurious or aggressive thoughts were experienced as non-anxiety-related. OCD-TD only, reported more washing behavior. Müller et al. (1997) reported echophenomena to be predictive of TS in 56% of cases when compared to OCD. Cath, Spinhoven, Hoogduin et al. (2001) found that the presence of touching behaviors accurately predicted diagnosis of TS in 77% of cases and echophenomena predicted 83% of TS. OCD and TS may have similar thoughts but TS show more playfulness with their thoughts and OC show more anxiety. Shapiro, Shapiro, Young and Feinberg (1988) have argued that repetitive thoughts and actions in TS are performed automatically as a consequence of a failure to restrain impulses and are not anxiety-related. A questionnaire designed by George, Trimble, Ring, Sallee and Robertson (1993) explicitly distinguishes the sensory-based overt and covert rituals of TS from the more intentional obsessions of OCD. Cath, Spinhoven, van Woerkom et al. (2001) also note the importance of distinguishing cognitive tics from mental obsessions. Cognitive tics are playful and usually aimed at tension reduction rather than harm avoidance.

2.3. Measuring tic behavior

There seem four preferred types of tic assessment: video rating, self-monitoring, clinician, and external rater assessment.

2.3.1. Video assessment

There do exist standard video rating procedures (e.g., the Rush method [Goetz, Tanner, Wilson, & Shannon, 1987]). One obvious problem biasing video assessment is that tics are frequently suppressed, so simply videoing a person may not be a reliable way of identifying the presence of tics, unless a tic-provoking situation is constructed in collaboration with the person. For
example, in a recent study in our laboratory, expert raters showed a poor hit rate in distinguishing tic from non-tic disordered clients during a standard videod psychiatric interview, but with nonetheless a good inter-rater agreement (O’Connor et al., submitted). But tic detection improves substantially during the video of a high risk situation.

2.3.2. Self-monitoring

The status of the tic or habit and associated emotions can be monitored in a daily tic diary which records frequency, intensity of urge to tic and degree of control (resistance) over the tic on a daily basis in a specially prepared booklet (printed prototypes are available on request from the author). Participants must be trained in the use of the booklet and a unit of tic (tics) or habit defined at the beginning of the evaluation.

The way the person appraises both the tic occurrence, appearance, and the long-term effects of suffering from tic disorder have not been systematically studied, It is possible that an appraisal–assumption instrument such as the OBQ-87 (Obsessive Beliefs Questionnaire [OC Working Group, 1997]) could be adapted for this purpose (Anholt, Emmelkamp, & Cath, 2002). Alternatively, O’Connor, Gareau and Blowers (1994) have suggested modifying the Kelly’s repertory grid to permit assessment of constructs defining reactions to high and low risk situations and to self as tic disordered (Blowers & O’Connor, 1996).

2.3.3. Clinician or external observer assessment

Here the observer or clinician rates the tic occurrence without the person’s knowledge. This is useful for overall improvement ratings. Unfortunately, identification of tics will be limited unless the contextual nature and suppression of tics is taken into account.

Each of these four measures of tic behaviors has a different type of bias, but agreement between all four assessment measures on outcome gives some convergent validity to a finding of a clinically significant change, or not, on the triangulation principle adopted in other types of clinical methodology (Kay, Guernsey de Zapien, Altamirano Wilson & Yoder, 1993).

3. Aetiology: neurobiological approaches

3.1. Neurophysiological factors

Current clinician consensus is that TS and by implication TD is a neurobiological disorder (although such consensus emphasizes equally the clinical importance of addressing psychosocial consequences; e.g., Leckman & Cohen, 1999). Essentially, the neurobiological model holds that TS/TD is inherited, probably by an autosomal dominant gene tic transmission (although recent gene mapping suggests it may be more complex and heterogenous [Pauls, 2001]). The trait inherited is described either as a difficulty inhibiting sensory urges, or as a difficulty inhibiting behavior due to problems in neurochemical modulation affecting cortical–subcortical areas. Although there does seem evidence of family pedigree, the criteria for inclusion in pedigree studies is often subsyndromal and there are other methodological problems with these studies (Fallon & Schwab-Stone, 1992). In any case, there is no firm evidence of any uniform organic deficit in TS or TD. In fact, neurobiological findings are very patchy, incoherent and as in other
psychiatric areas, amenable to alternative behavioral interpretations. For example the role of tic behavior itself and associated coping behavior (e.g., presence of tic suppression) in producing changes in brain functioning is little explored.

The principal circuit hypothesized to be implicated in tic production is the cortico-striato-thalamo-cortical circuit. This includes thalamus, basal ganglia, motor areas and orbital-frontal cortex. These are essentially the areas involved in controlling all movements, so it would not be surprising, if TS/TD is hypothesized to be a movement disorder, that these regions were involved. However, the exact fault in this circuitry remains in dispute. One hypothesis, that of a structural deficit, is mostly derived from lesion studies and from studies of actual basal ganglia dysfunction which produce motor impairment (see Peterson, Leckman, Arnsten, Anderson, Staib, Gore et al., 1999). Parallels are constantly drawn between tic disorders and basal ganglia dysfunction disorders such as Huntington’s chorea, but there is no firm evidence of common structural deficit. Another hypothesis, that tics result from increased motor neuron firing which induces clonus-like activity (e.g. Smith & Lees, 1989) remains speculative, and anyway lacks specificity to TS/TD. An alternative speculation that tic behavior (and OCD) may result from an abnormality in the neural basis of innate motor routines (e.g. Knowlton, Mangels, & Squire, 1996) relies overly on linking tics and rituals to species specific innate behavior.

There is, however, some consistent evidence, from neuropsychological studies in children with TS, of problems with motor skills and in particular visuo-motor integration (Schultz, Carter, Schall, & Leckman, 1999). Here though the child populations studied frequently had comorbid ADHD. Such neuropsychological tests have reported abnormalities in severe TS in skilled motor tasks like the Purdue pegboard and Groove tasks in children (Hagin, Beecher, Pagano, & Kreeger, 1982; Bornstein, Baker, Bazylewich, & Douglass, 1991), pre-adolescents (Bornstein, 1990) and adults (Bornstein, 1991). There is also robust evidence of a higher level of overall motor activation in TD/TS. This activation takes the form of a higher cortical arousal, and higher behavioral arousal, as shown in problems inhibiting automated motor actions (e.g. O’Connor, Robert, Dubord, & Stip, 2000; O’Connor, Lavoie et al., 2001) and restless legs (Leckman & Cohen, 1999).

More direct evidence, based on functional magnetic resonance imaging (fMRI) studies (Biswal, Ulmer, Kripendorf, Harsch, Daniels, Hyde et al., 1998), has shown an overactivation of the sensorimotor and supplementary motor area and recruitment of larger portions of these areas in five patients with TS during the execution of a finger tapping task. O’Connor, Serawaty and Stip (1999) and O’Connor et al. (2000), using a countermanding paradigm adapted to be an analogue of a high-risk tic situation, found no difference in psychomotor speed between TD and non-tic controls, but the authors did find that participants with TD showed more difficulty when inhibiting an automated than a controlled response and showed no practice effect in performance over trial blocks. Ziemann, Paulus and Rothenberger (1997) reported evidence of normal motor threshold and excitability but reduced or impaired motor inhibition in TS. The finding relates to inhibition but not to excitability, and likewise Georgiou, Bradshaw, Phillips, Cunnington and Rogers (1997) found no impairment in TS in fast, goal-directed movements.

These motor inhibition problems may themselves, however, be due to strategy rather than deficit. For example, distraction from tics, or hyperactivity or tic suppression could affect performance (Channon, Flynn, & Robertson, 1992; Silverstein, Como, Palumbo, West et al., 1995). Motor differences between adult TS/TD and controls, for example on the Purdue Pegboard, can decrease following behavioral treatment (O’Connor, Brault, Robillard et al., 2001). Intriguing evidence
from recent brain imaging studies suggests that cortical functional differences may result from functional compensation of the brain to tic suppression. A recent plausible hypothesis, in this vein, is that the need to suppress tics produces striatal hypermetabolism, and that compensatory mechanisms may even lead to larger orbito-frontal and parietal brain adaptations (Peterson et al., 1999; Peterson, 2001). But longitudinal studies are necessary to establish this link.

3.2. Neurobiological treatments

Hypothetically, both TS and TD could involve dysregulation of the dopamine (DA) system. Elevated DA could be consistent with the state of heightened activation, noted above, in TS (Muller-Vahl, Berding, Brucke, Kolbe, Meyer, Hundeshagen et al., 2000). The hypothesis of elevated DA in TS/TD would seem plausible given the presence of DA receptors, in projections to frontal areas, so supporting the link between DA and motor activation. DA neurons are also found in mid brain structures such as the substantia nigra and the system projects to a variety of brain structures including frontal areas. Hence, the DA hypothesis could be concordant with a brain imaging study which found that TS showed significantly elevated right frontal activity compared with controls (George et al., 1992). There is, however, no difference between controls and TD/TS in baseline DA levels or DA turnover (see Anderson, Leckman, & Cohen, 1999 for review, p. 266), which has led investigators to propose more elaborate DA deficits such as disturbance in the balance of D₁ to D₂ receptors and differences in the number of DA transport sites. Observations consistent with a hyperfunctional DA system come principally from the limited success of treatment protocols using DA antagonists. But clinically speaking, not all patients improve as a result of neuroleptic administration (Regeur, Pakkenberg, Fog, & Pakkenberg, 1986).

The initial drug of choice for TS, a low dosage of haloperidol, had the potential for unwanted extrapyramidal effects and noradrenergic side effects. Consequently pimozide is now preferred in terms of efficacy and side effect profile and shows good clinical response in some studies, but is contested in others (Sallee, Nesbitt, Jackson, Sine, & Sethuraman, 1997). Pimozide antagonizes DA receptors with selectivity for the D₁ dopamine subtype. Other dopamine agents antagonistic to presynaptic D₂ receptors similar to pimozide such as pergolide have shown benefit in TS (Griesemer, 1997). However, some of these dopamine agents show “tardive Tourette’s” (i.e. paradoxical worsening of TS), secondary to neuroleptics. Atypical neuroleptics such as olanzapine have shown more favourable outcome and fewer side effects in comparison to the more typical neuroleptics such as pimozide (Robertson & Stern, 2000). However, DA agonists such as deprenyl and clonidine have also shown efficacy in TS and alpha-2 adrenergic agonists such as quinacine have reported improvement (Chapell, Riddle, Scahill, Lynch, Schultz, Arnsten et al., 1995). On the other hand, stimulants such as methylphenidate (MPH) or dextroamphetamine are controversial, often showing initial worsening of motor tics whilst improving ADHD and disruptive behavior. However, recent evidence suggests that eventually after 18 weeks, even stimulants may lead to improvement and a combination of MPH + clonidine may give the optimal effect (Kurlan, 2001). Butolin Toxin (Botox) which essentially impairs functional innervation of the muscle leading to temporary atrophy has been reported useful in some cases of vocal tics (Jankovic, 1997; Scott & Jankovic, 1996), but in other case studies, the urge to tic was not reduced and the tic was more often than not replaced with another tic (Carpenter, Leckman, Scahill, & McDougle, 1999, p. 380). Other pharmacotherapies which in single cases seem to have produced temporary
relief from tics include: cannabinoids, nicotine: opioids, lithium, benzodiazepines, calcium antagonists, hormone therapy. The neurochemical picture becomes more complex in the light of differences between TS/TD and controls in other neuro-transmitter systems such as GABA, serotonin and glutamate (Anderson, Leckman & Cohen, 1999). Serotonin depletion and dopamine excess may be part of a dual mechanism regulating activation. Serotonin has a larger cortical concentration in frontal areas and has been linked with motivational and mood factors, in particular during stress, whereas DA may be more involved in reward and stimulation. So effectively the neurochemistry of tic disorders may implicate both cortical and sub-cortical neurochemical factors, and hence both excitatory and inhibitory motor processes. This may explain the paradox of tic behaviours, exhibiting characteristics of disinhibition in their impulsive reflex-like nature, and of inhibition in the form of resisting or suppressing activity at the same time. Cath, Spinhoven, Landman et al. (2001) suggest that TS spectrum as opposed to OC spectrum are less responsive to selective serotonin reuptake inhibitors (SSRI) and more responsive to selective noradrenaline reuptake inhibitors (SNRI).

Apart from serious side effects, response to neurobiological treatment regimes is variable, and the tics themselves are rarely eliminated by neuroleptics or other types of medication (Regeur, Pakkenberg, Fog & Pakkenberg, 1986). Double-blind placebo-controlled designs have found tic frequencies reduced by about 50% using haloperidol or pimozide (Shapiro, Shapiro, Fulop, Hubbard, Mandeli et al., 1989) and unwanted side effects occur in about 80% of individuals. Also, only about 20–30% continue their medication for an extended period of time (Peterson & Azrin, 1992). New medications continue to appear on the market not exclusively tailored to tics, but rather generalized from other neurological problems such as Sydenham or Huntingdon’s chorea. A number of single cases have reported diverse stereotactic surgical procedures with beneficial results including: limbic and frontal leucotomies, bilateral cerebellar dentotomy, ventro-lateral thalamotomy and gamma capsulotomy. But long-term outcome in these surgical cases is not well documented (Rauch, Beer, Cosgrove, & Jenike, 1995).

In summary, the neurobiological and neurochemical chemical findings are puzzling and inconclusive, with practically every neurotransmitter system seemingly involved in at least one successful case study treatment of TD/TS. This includes medications which are both agonist and antagonistic to a DA model (e.g. MPH) and even within antagonists there are controversies over the comparative effectiveness of some neuroleptics (e.g. Onofrj, Paci, D’Andreamatteo & Toma, 2000).

4. Aetiology: behavioral approaches

4.1. Learning model

An alternative approach to tic development is found in the learning model initially proposed by Azrin and Nunn (1973) which views tics as learned responses or, more specifically, adapted startle reflexes. A traumatic event evokes a reflex which develops into a tic which is then maintained by self-reinforcing factors (see Fig. 2). Likewise, according to Commander, Corbett, Prendergast and Ridley (1991), the tic is a form of startle reflex that is learned in response to an aversive event, although, according to these authors, the propensity to be startled and overstimul-
Injury / Illness / Stress / Critical events elicits a startle response

Startle reflex motor response / Defensive motor reaction relieves aversive sensation and or muscle tension

Startle response reinforced internally/externally by adjunctive/intermittent reinforcement schedule

Lack of control/awareness of learned response facilitates auto-reinforcement and automated adoption of response

Startle response develops into over-learned automated tic habit

Fig. 2. Schema of the learning model of tic development (adapted from Azrin & Nunn, 1973; Commander & Cobett, 1991).

Azrin and Nunn (1977) hypothesized that tics develop following some physical trauma because they relieve muscle tension resulting from the injury or in other ways protect from injury. Tics are then negatively reinforced by the tension reduction that follows the occurrence of the tic behavior (Evers & Van De Wetering, 1994), or by other external factors such as social attention. Additional contingencies encourage the propagation of the tic. For example, since the person lacks awareness of the tic, s/he can never exert complete control over the tic. Also, the tic may occur as an adjunct to other self-reinforcing behavior on an intermittent schedule of reinforcement, which leads the tic to become an auto-reinforced over-learned habit, difficult to extinguish.

Evidence supporting the early developmental part of the learning model is patchy. Sachdev,
Chee and Aniss (1997) found no evidence of abnormal audiogenic startle reflex in 15 TS compared to 15 controls using stimuli at 88 and 114 db. But more recently Gironell, Rodriguez-Fornells, Kulisevsky, Pascual, Riba et al. (2000) did report on exaggerated acoustic startle reflex in 10 TS compared to 10 controls presenting 110 db signals in a start-react paradigm. There is evidence of tics developing subsequent to peripheral physical injuries and Factor and Molho (1997) report two such cases. Tijssen, Brown, Morris and Lees (1999) reported three late onset startle-induced tics, two linked to physical trauma and one linked to emotional stress. Some tics can be traced to learned gestures (e.g., scratching developing after a bout of acne, blinking after an eye operation, a head tic beginning due to the long hair worn during adolescence). This information, clinically useful though it may be in helping the person understand the morphology of the tic, proves nothing about the learned nature of tics because such a situation may simply have been the occasion for the tic developing. In TS in particular, tics can wax and wane, and be adopted on the basis of suggestion as an echo phenomenon (Seligman, 1991).

The claim that tics result from trauma or injury then has some support, but not every tic develops subsequent to startle or injury, and the injury hypothesis does not account for tic substitution where a tic in one part of the body may substitute for, or replace, a tic in a distant part. Also, the precise reinforcer for the tic habit is unclear within the learning model (Turpin, 1983). Does the person engage in the tic habit to stimulate the self, or to rid the self of unpleasant sensations. In other words, is the tic reinforced by positive (stimulation) or negative (discomfort avoidance) reinforcement.

The tic habit is most likely reinforced through tension relief (Evers & Van De Wetering, 1994). However, any relief is short lived. Physiologically, the tic takes the form of a series of tense-relax muscle cycles in an already tense muscle where, however, the relax phase does not reduce tension to zero, but effectively returns it to an already elevated baseline. The long-term result of ticcing is hence maintained or increased muscle tension. The tense release cycle could be seen to function in a way similar to rituals and other neutralizations in obsessional and habit disorders, which subjectively comfort whilst augmenting or maintaining the problem in the long run.

However, the tense/release cycle maintenance model of tic behavior is contested by another psychophysiological model emphasizing heightened sensory awareness as the maintaining factor in TS/TD (Fahn, 1993). This model proposes that tics represent a reaction to a heightened sensitivity towards sensory signals or alternatively an intolerance of such sensations. This heightened awareness model principally challenges the assumed involuntary nature of ticcing (implicit in the tension/release model). Is the tic, as Hollander (1993) suggested, a sign of lack of control over motor inhibition? Or is it the sensory, premonitory urge or sensation which is involuntary and which leads to the tic action? Bliss (1980) also suggested that it is the premonitory urge to tic which is compelling, not the action. The tic action then constitutes a (semi)-voluntary response to the premonitory urge (semi-voluntary since the tic is still an automated not a planned action). If the tic is a (semi)-voluntary response to cope with the hypersensitivity, then the preferred treatment strategy for tic management would be exposure and habituation to the premonitory sensory urge as an alternative to reversing the motor tic habit through habit reversal (Bullen & Hemsley, 1983; Hoogduin, Verdellen, & Cath, 1997).

But this latter treatment approach begs the question: what exactly is the premonitory sensation or urge? Its nature seems at the same time sensorial and attentional, and it is difficult phenomenologically to distinguish the two aspects (see Fig. 3). The sensation is usually considered a sensory
tic, indicating tension in the surrounding area. It is not clear if this sensation serves as a warning, a precipitator, or is in fact part of the tic, because the sensation can persist even when treatment eliminates the actual tic movement. One possible interpretation of premonitory sensory tics is that they represent the subjectively experienced component of neural dysfunction below the threshold for motor or vocal tic production (Chee & Sachdev, 1997). Kane (1994) has suggested that premonitory urges represent a heightened attention to physical sensations. He suggests that a particularly heightened sensitivity of the person with tics to somatic sensation produces an attentional focus that provokes the tic. If heightened awareness or attentional hypersensitivity to a particular tension becomes a preoccupation, and the attempt to suppress such preoccupation provokes a tic, then this tic-producing process resembles the thought suppression analogue of obsessional thought patterns, where the attempt to suppress an intrusive thought results in its resurgence (Purdon, 1999). Although the premonitory urge clearly correlates with tic frequency, it does not precede
every tic, it can arise independently, can persist when the tic behavior is eliminated, and can be modified independently by distraction and habituation (O’Connor, 2001).

The premonitory urge may have similarities to the craving to smoke, which although seemingly inciting the smoking behavior actually varies independently of smoking and has a cognitive component (Tiffany, 1990). In the case of cigarette craving, the decision to smoke or the anticipation of smoking can produce the craving (O’Connor & Lanlois, 1998). Possibly then the premonitory urge may be a product rather than a cause of attentional focus and may constitute an anticipation of ticcing subsequently reinforced by the ensuing tic behavior rather than an independent sensory precursor (see Fig. 3).

4.2. Behavioral treatments

The learning model has produced the most compelling behavioral treatment for tics to date: habit reversal (HR). A great many behavioral treatment techniques have shown some success with tic management, including relaxation, hypnotism, muscle feedback, awareness training, negative self practice, response prevention and massed practice (see Azrin & Peterson, 1988), but the most impressive results have been reported by Azrin and co-workers using HR. Using this method, Azrin & Peterson, 1988, 1989, 1990) and Peterson and Azrin (1992, 1993; Peterson, Campise, & Azrin, 1994) report a reduction in tic frequency in the home environment of up to 99% in several studies involving both TD, HD and TS. The HR package involves multiple stages, including relaxation, awareness, contingency training for positive reinforcement of non-ticcing and training in a competitive antagonistic response. This latter technique involves tensing the muscle antithetical and incompatible with the tic-implicated muscle. Awareness training and competing response training seem the crucial elements of the program (Miltenberger, Fuqua, & McKinley, 1985).

The competing response (CR) theoretically installs an alternative response or habit to counter the tic movement and hence eliminate it. But there are alternative accounts of the CR. For example, the CR may function as a “geste antagonistic” whereby a random movement or reaction in a different part of the body by itself modifies the tic or habit. The CR might function as a movement distraction. Equally the CR may function as a punishment for habit onset (Turpin, 1983). It may function as a negative or positive reinforcer or as a social reinforcer, or as automatic reinforcer, or by some as yet to be identified means. Alternatively the CR may be effective as a class of schedule-induced or adjunctive behavior occurring as a side effect of other time-based reinforcement schedules. In other words, the tics/habits may simply be performed when the person has time on their side and can easily be replaced by the CR (Woods & Miltenberger, 1995).

Although HR shows good response in isolated cases and seems to be equally effective in TD, TS and HD, its psychosocial impact and generalization is not clear. Unfortunately Azrin and coworkers do not give sufficient information on the psychiatric co-morbidity of their sample to permit adequate sample comparisons. Also, it is clear that the latter’s criteria of success is limited to tic prevention and does not measure the impact on other behavioral or psychosocial factors. Studies of HR continue to show useful gains in clinical practice (Wilhelm, Deckersbach, & Coffey, 2001; O’Connor et al. 1997; O’Connor, Brault et al., 2001) reported successful outcome of the HR package in the treatment of TD. Clinician consensus, however, as voiced at a recent clinical round table discussion at the Tourette Syndrome Foundation of Canada in Toronto (2001) is that HR is generally ineffective as a routine treatment strategy in TS. Clearly more studies
aimed at elaborating the theoretical basis of HR, standardizing application and establishing generalizability of findings across tic subtypes would be helpful. An alternative treatment approach, as noted previously, is exposure to the premonitory urge during response prevention of the tic. This treatment approach is similar to the exposure and response prevention treatment model in other problems where exposure to the anxiety-provoking obsession induces habituation and reduces the need to neutralize the sensation (i.e. to tic). Hoogduin, Verdellen and Cath (1997) presented four cases showing limited success and suggest this approach may be particularly suitable for severe cases.

5. A cognitive–behavioral/psychophysiological model

The current cognitive–psychophysiological (CP) model, whilst building on previous behavioral research into the development, maintenance and modification of learned habits, proposes that cognitive factors play a much larger role in both the emergence and the maintenance of tic behavior, and that both the development and modification of tic behavior is best understood in terms of a reciprocal interaction between psychological and physiological factors. The model has been developed over the last eight years, working mainly with simple and complex chronic TD adults as well as HD and mild-moderate TS populations. Its generalizability to more severe TS is discussed later. Fig. 4 provides a schema of the interacting components of the model. Fig. 5 more explicitly discusses behavioral strategies related to the emergence of any specific tic cycle. Fig. 6 illustrates the target components for reducing motor activation and associated tension, as proposed in the model.

The CP model in Fig. 4 describes background factors contributing to the onset of current tic behavior. The model is mute concerning the more distant influence of genetic transmission or associated inherited pathophysiological dispositions. The window of the model is the “here and now” maintaining current tic behavior. The directional flow is not strictly causal, but rather illustrates the reciprocal influence of successive levels of background factors on tic development. The model postulates two independent source vectors, one cognitive/emotive, the other behavioral/physiological, but leaves open the possibility that these two sources may themselves have distinct aetiological roots. However, a key determinant, according to the CP model of tic onset, is precisely the reciprocity in everyday practice between cognitive/emotive and behavioral/physiological factors. The metaphors of depth and distance may be appropriate to apply to the organization of the reciprocal-factors at successive levels of the model. Each successive level, working back from tic onset, represents the ground from which factors at the preceding level emerge. However, the starting point of the model is at both a cognitive and behavioral/psychophysiological level. There is a cognitive style concerning the right way to act, organize and plan activities, on the one hand, coupled with a heightened self-attention and sensory focus which also functions at the psychophysiological level. The starting point from the cognitive side is a perfectionist style of planning action. The perfectionism relates most closely to the personal organization and the personal standards subscales of the Frost Multi-dimensional Perfectionist Scale (MPS) (Frost, Marten, Lahart, & Rosenblate, 1990). On the behavioral/physiological side, there is heightened sensory focus, hyper-sensitivity and heightened activity. All elements of the model are already established attributes of TS/TD. The model, as elaborated in the following sections, offers an account of how the reciprocity between elements influences tic onset.
5.1. Perfectionist style of action

People with TD, TS and to a lesser extent HD, seem to score higher than controls on subscales of the MPS related to personal standards and organization. However, TD do not necessarily score high overall on measures of perfectionism or obsessionality (O’Connor, Brault, Loiselle et al., 2001). The cognitive aspect of this perfectionism manifests itself in beliefs about the importance of being efficient, of doing as much as possible, and not wasting time, or appearing to do
so. These perfectionist beliefs are accompanied by a premeditated style of action involving a tendency to attempt too much at once, premature abandonment of tasks, unwillingness to relax, pace action appropriately, invest more effort than necessary, be in advance of self and foresee the unforeseeable. This is a finding so far replicated in two separate studies (O’Connor, Gareau, & Borgeat, 1997; O’Connor, Brault, Loiselle et al., 2001).

The behavioral aspects of this style of planning action first came to notice in the clinic when clients couldn’t find time to do the homework exercises because they already had too much to do. It would be tempting to connect this planning style with ADHD in terms of impulsivity, distraction, hyperactivity or even with type A behavior. This over-active style clearly bears a
“family resemblance” both to hyperactivity and possibly “type A” behavior. As Leckman and Cohen (1999, p. 71) note, there are few studies assessing hyperactivity in adults. Hicks, Conti and Bragg (1990) have previously reported increased type A behavior in HD. However, the term “overactivity” is preferred here since it avoids assumptions of a link to childhood hyperactivity and grounds the description in the precise behavioral observation in the adult of over-investment in planning action. The style of planning here is clearly premeditated and aimed at achieving a perfectionist goal. Although the style of action may be idiosyncratic and situation-specific to the person, the perfectionist appraisals are uniform and persistent and represent a stable belief in the overactive style as the most efficient way of acting. The person with TD may report rigid black and white beliefs such as: “Either I do everything at once or I’m lazy”. Or there may be unspoken rules and assumptions which make the person feel they must act in this way, e.g. “Not feeling well is not a good enough excuse to change a scheduled appointment/activity.” “People will not tolerate me if I’m a little late.” There may also be deep core assumptions fuelling overactivity such as “I could be inadequate and if I don’t act as quickly as possible, everyone will know I can’t perform” Such a perfectionist style of action becomes self-perpetuating.

These observations on “overactivity” were recently incorporated into a formal 30-item style of action questionnaire (STOP) whose subscales discriminated well (74%) between TD, OCD and controls. Two robust dimensions emerged from the factor analysis (n=260): over-activity and over-investment in preparation. Internal consistency and test-retest reliability of high loading items for the tic sample were good (O’Connor, Aardema, & Brisebois, 2001). The first dimension reflected continual activity, a difficulty to keep still or to do one thing at a time. Typically, the overactive person will have several tasks planned at the same period time (ex. go to the mall, ...
return library book, visit a friend, go to the bank, etc.). The result, apart from always being in a rush, is that the person feels stretched and strained and constantly in conflict between what s/he is doing and what s/he feels should be done. Frequently this conflict adds in an unnecessary stress to current tasks and sabotages feelings of accomplishment. The offshoot is that the person is dissatisfied, frustrated, irritable, feels trapped, and judges the self badly. The second dimension, over-investment, relates to doing more than is necessary, and expending more effort than necessary on an intellectual, emotional and physical level. On a physical level, the person may invest irrelevant tension in another part of the body or in the same muscle group, for example blinking with cheek or forehead muscles in addition to eyelid muscles, or respond to an isolated task with a block movement (lift up a pen with the complete arm, not just the fingers). The tic action itself is often visible precisely due to the implication of non-pertinent muscles. This extra physical effort can be normalized with practice but there may also be a tendency to over-invest intellectually and emotionally in the reaction to, and anticipation of an event. Even if the person is immobile during the tic, thoughts can create unnecessary frustration and an over-investment in negative anticipation and preparation (O’Connor et al., 1994).

5.2. Heightened sensory awareness

The hypersensibility of TS/TD has been noted by several authors in visual, auditory and tactile modalities (e.g. Bliss, 1980; Kane, 1994). Such sensitivity can lead directly to the development of tic-like responses in coping. For example, as noted, eye blinks may originate as defensive responses to light. Subjectively TS/TD report increased sensory discomfort, tingling, or itches, sometimes resulting directly in scratching, rubbing or moving (e.g. Leckman and Cohen, 1999, p. 27). As noted earlier, the premonitory urge prior to ticcing could itself be a result of such hypersensitivity, and there is some experimental evidence of sensory excitability and augmented sensory evoked potentials in TS/TD (van Woerkom, Roos, & van Dijk, 1994; Johannes et al., 2001). However, there is a potentially cognitive aspect to this sensory element, albeit relatively under explored. Increased attentional focus might augment hypersensibility to sensations, and the possible attributions of negative significance to the sensations may further increase their importance in a similar way to the cognitive distortions and appraisal assumptions in other psychiatric disorders (ex. panic, somatoform disorders) (Salkovskis & Clark, 1993). In addition, enhanced self-attention and self-awareness may result from increased self-focus. A fairly consistent finding with TS/TD is the sensitivity of such clients to the judgement of others (Leckman and Cohen, 1999, p. 149). The person with TD is over-concerned with self-image, that others will pass judgment (including on appearance of the tic), and detect subtle deficits in reaction and style, leaving the person dissatisfied with him/herself. In a recent study, items of self-image distinguishing TD from controls were mainly concerned with feelings of being ill, at ease with others, and dissatisfied with self-image (O’Connor, Brault, Loiselle et al., 2001). Although such self-focus and sensitivity could, of course, result from the experience of ticcing through conditioned evaluation and subsequent generalization, the person’s awareness of the tic and its social effect is sometimes limited and the self focus can be present even in situations at low risk for ticking.

So there could be a reciprocal influence between the two initial cognitive/emotive and behavioral/psychophysiological starting positions. A perfectionist concern with personal standards could lead to a heightened self-attention, sensory awareness and a self-focus on appearance. Like-
wise, a heightened hypersensorial state could lead to heightened agitation and an over-concern to regulate personal actions and appearance. Both positions independently would in any case ultimately enhance sensory stimulation and feed sensory–motor activation. A high level of motor activation has been documented in TS/TD (see previous section). There is evidence of motor neuron excitability, enhanced motor preparation, difficulty inhibiting responses or modulating arousal when initiating complex responses. Enhanced motor activation would in turn affect neurochemical regulation which could in turn produce increased motor excess and restlessness, and further encourage motor activity, impulsivity and perfectionist concerns. Cath, Spinhoven, Landman, and van Kempen (2001) in a study examining serotonin activity in TD and OCD, have suggested that low serotonin syndrome and 5-HT hypo-functionality is associated with impulsivity. The complex relation between, say, serotonin and DA may not, in this case, be pathologiespecific. Hence differences in neurochemical levels may be less a state function and more a long-term compensation/adaptation mechanism in response to chronic dysregulation of activity and arousal.

This reciprocal path between neurochemical and sensori-motor activation leads at the same time to both greater stimulation, and the need for more such movement and stimulation. This in turn would feed sensory–motor activity and result in a difficulty to remain calm. Motor activation would also be sustained directly by continued beliefs about the need always to be on the move. All these influences could produce a ceiling level of motor activation or “motor excess” (Tryon, 1993).

5.3. Dysregulation of arousal, problems in visuo-motor integration, and reliance on somato-sensory proprioceptive feedback

This ceiling effect, due to chronic over-activation, could plausibly lead to problems in short-term arousal regulation. But this arousal regulation could also be disrupted by distraction from the perfectionist desire to be always ahead of oneself. The latter tendency could lead to less attentional focus on performance in the here and now, with thoughts focussed more on the next task or on future, or other, events. Anecdotally, TD clients frequently complain of being “in the moon” and not being entirely focussed on the here and now. Sample quotes: “I’m always ahead of myself; I’m doing one task and already I’m daydreaming about what I must do next and so on.” “I’m rarely reacting in the here and now, to the present.” “I find it difficult to not think on ahead. There always needs to be something going on in the back of my head.” There may be difficulty in optimally adapting arousal/activation level to task demand. Because of the emphasis on feeding sensory–motor stimulation, external information indicating when tasks are satisfactorily done, or the appropriate goal for a task, may be trumped by information from proprioceptive feedback, rather than adjusted in accordance with complex integration of current visuo-spatial information. This reliance on intensity of muscle tension and effort may resemble the proprioceptive focus typical of somatoform disorder (Scholz, Ott, & Sarnoch, 2001).

An overactive trying-to-do-too-much-at-once, and over-investing-too-much-effort style of action could well impair efficient regulation of arousal and attention. Difficulties in regulating attention/arousal would result in, and be, in turn, reinforced by restlessness. Such difficulties in regulating arousal and attention could explain the specific problems of TD/TS in initiating and carrying through complex tasks (which require controlled allocation of resources). Problems in initiating complex tasks could lead to a level of frustration associated both with the consequences
and with the anticipation of complex task performance, particularly those tests requiring open loop planning (i.e., controlled rather than automated regulation). This frustration might also impair the TD/TS ability and/or desire to plan actions on the basis of visuo-spatial cues and so integrate these into movement planning. Problems in visuo-motor integration (noted earlier) could thus result from an over-reliance on somato-sensory and proprioceptive “feel” to know when the action is accomplished or when enough effort has been invested. The frustration and impatience would then lead to enhanced tension as the muscles are continually over-readied for action (action–frustration cycle) and kept in a state of over-preparation relying on somesthetic and proprioceptive (rather than visual) information on accomplishment.

5.4. Chronic muscle tension

Obviously, by definition, a muscle must be tense in order to tic. The tic implicated muscle is frequently a muscle implicated in a task where the action–frustration cycle is most notable and recurrent. Electromyographic (EMG) recordings of tic-affected muscles show that these muscles are rarely associated with zero contraction and have a greater difficulty compared to non-affected muscles in achieving different degrees of contraction rather than an all or nothing state of tension (O’Connor et al., 1995). People suffering from tics do also subjectively report chronic tension. Hoogduin et al. (1997) reported high overall muscle tension as a consistent feeling in all patients when identifying premonitory urges.

So how would such muscular tension be built up prior to tic onset? Behaviorally speaking, tension may be a product of inappropriate over-investment in action, or disproportionate anticipatory activation of the muscle at a premature stage in motor execution. On rare occasions, the tic may be traceable to a learnt action particularly in TD, or an accelerated series of actions learnt in reaction to stress or other related situations. In a recent electrophysiological study (O’Connor, Lavoie et al., 2001) looking at electro-cortical preparation and movement execution potentials, participants with TD showed no consistent relationship between the preparation and execution stages of action, suggesting that the preparation stages were not modulated efficiently, due to a high ceiling level of activation. Anticipation and preparation can themselves induce muscle activation through the influence of the gamma-efferent system, and clients report that anticipation of a tic and focus on the tic muscle can by itself produce the tic (Kane, 1994). Even purely intellectual recall of a past tic can reactivate it (Peterson et al., 1999, p. 257). High tension generally would be maintained by over-preparation, which would not permit the muscle to profit from a normal relaxation cycle. The tic cycle is a series of tense–release contractions whose goal is to release, temporarily, tension or alleviate an aversive sensation, but which effectively maintains tension. The tension release cycle keeps the muscle in a chronic state of preparation to act, and this chronic state in turn simulates a state of readiness (O’Connor, 1989). Consequently, progressive muscle relaxation as a therapeutic strategy seems beneficial and forms an important component of HR treatment. The CP model for understanding the interaction of tic onset and background tension and the different inputs into chronically high tension is given in Fig. 6. All of these inputs contribute to an increased level of sensory–motor arousal. The differential contribution of each of these inputs needs to be considered to reduce tension. A behavioral analogue of a high-risk tic situation of over-preparation is scripted in Table 1.
You are preparing an action with a specific set of muscles in order to carry out a complex task with several unknowns. You are concerned to be judged well by others for your performance (e.g. skilled spectator sport, a spontaneous public speech, an exam situation). You wish to get the task over with as soon as possible and feel you should always be further ahead in your performance. Just as you are about to start the action, you must hold the action but be ready to restart it again at an unspecified time in the future.

Result: Chronic over-preparation accompanied by frustrated-action, followed by a repetitive tense–relax cycle in task-related muscle groups.

5.5. Feelings of frustration associated with tic onset

The tic occurrence is almost always accompanied by feelings of frustration or dissatisfaction. O’Connor et al. (1994) looked at situations likely to evoke tics, and found these were not necessarily related to anxiety but rather to thoughts or events of frustration. In particular low-risk tic perceptions of “feeling free to act” or “being myself” were opposed to high-risk feelings of constraint, obligation, and being judged by others.

Tic occurrence has a situation/activity profile. In other words, the tic is more likely to appear in some rather than other situations or activities, or is more likely to be worse during some situations or activities than others. The high risk situations/activities may be, objectively speaking, very idiosyncratic and puzzling. One person tics when reading, another when out walking. Despite the personalized nature of high-risk situations/activities, appraisals of such activities tend to be more uniform. During such situations/activities the person feels dissatisfied, bored, frustrated, and under pressure. A more recent study (O’Connor, Brisebois, Brault, Robillard, & Loiselle, 2002) confirmed that tic onset is predominantly associated with dissatisfying and tension-producing activities.

The CP model proposes that tics always occur in situations which represent to the person the height of dissatisfaction or action-frustration. Being late for an appointment, for example, by itself may not be enough to evoke a tic, but being, in addition, late and stuck in the traffic and anticipating a stressful interview may cumulatively elicit the tic. The tic is like the tip of an iceberg with a more general high level of background tension below the surface, and as we discuss later, according to the CP model, it is necessary in treatment to reduce the tension and activation level overall as part of behavioral restructuring as well as addressing the tic in isolation (see Figs. 5 and 6) in order to avoid tic substitution.

5.6. Tic onset and muscle use

According to the CP model, the tic onset occurs against a general background of tension. But why does the tic onset in one group of muscles and not others? In our clients, muscle implication in the tic seems ultimately linked to muscle usage. Although in TS tics can move around the body, this fact would still support the usage model as long as the tic occurs in muscles linked to an action–frustration cycle. Tics occur in otherwise voluntary muscles linked either by usage or expression to a high risk tic situation. For example, a masseuse develops a twitch in her most
forceful dominant hand when she is trying to rush a massage, a trombone player develops a tic in his lips, mouth and tongue during a hasty rehearsal, and more symbolically another person grimaces and makes a sound like “tsk tsk” with her mouth when anticipating problems. The sound and movement resemble an expression of negative self-judgement and frustration.

The tic then usually occurs in muscles linked either by symbolic or physical association with use, or preparation for use, in a high-risk tic onset situation. The tic-affected muscle might be implicated in an expression of emotion or action adapted for use in the high-risk situation or activity. For example, a client with an eye tic whose style of action involves always speaking forcefully and overinvolving face and cheek muscles in speech, feels he must fixate his addressee in order to not miss information in an encounter. He considers quick blinking an asset to help “use” in interpersonal encounters. So the tic action may serve part of a more general behavioral purpose and be linked to cognitive as well as physical goals. Obviously a defensive or startle reaction (as proposed by the learning model) would count as one example of such use, but such a reaction is only one category of use.

5.7. Coping strategies maintaining the tic cycle

The reaction to tic onset is also fed by a series of other cognitive and behavioral coping strategies (see Fig. 5). The person’s own model of tic development, management and consequences is an important influence on the anticipations and reactions to the tic. Clients may subscribe to a hydraulic model of tic management (ex. “If I keep it in, it will build up and I’ll have to let it out later one way or another since I can’t contain it.”) The person then adopts behavioral strategies which amplify this hydraulic sentiment and so confirm its validity by, for example, accepting the interpretation that detection of the premonitory urge means that a tic is imminent (Leckman, Walker, & Cohen, 1993, p. 102). As discussed earlier, it is not clear if this premonitory urge precedes or follows the anticipation of a tic, and hence whether the urge results from an attempt to deal with the tic, or whether it is an independent warning sign. Other appraisals relate to how the person feels the tic will be received and appraised by others in the situation. These appraisals may raise arousal and may explain how, in some cases, just the anticipation of ticcing can raise tension and frustration and hence, by itself, provoke a tic.

Behavioral strategies themselves adopted to cope with tic behavior (see Fig. 5)—divide into: containment, correction, concealment (or the 3 C’s). Containment would include tensing, holding in, or adopting a posture to contain the tic, such as laying down, or contracting the muscle to attempt an informal competing response. Correction includes actively occupying the muscle by integrating the tic into an action, or trying to normalize the tic action by modifying its course, or actively converting it into another movement. Concealment would include disguising the tic by wearing baggy clothes, carrying a bag over the shoulder, masking the tic through performing a larger act (e.g., sneezing), or finally by avoidance of tic-evoking situations. These gestures fulfill the same role as neutralization, avoidance or coping strategies in anxiety disorders and effectively reinforce apprehensions about ticcing and so in the long term, maintain the tension level.

5.8. Clinical implications

Clinical implications of the CP model go beyond current CBT treatment recommendations. The first implications are for the evaluation of tics. The model would suggest that in addition to
functional analysis, topography of the tic itself be studied in detail to give information not only on function but on usage. If the model of multi determined sensory—motor activation and chronic muscle tension production is correct then the evaluation should also seek out a profile of high-risk activity/situations and associated cognitive appraisals and coping strategies defining these high-risk activities. EMG evaluation would be a useful measure to evaluate objectively the level of muscle tension during ticcing. EMG could also form part of later biofeedback and training in aiding muscle discrimination aimed at reducing over-preparation and normalizing tic-related actions (see later).

Ticcing maintains tension. So a typical sequence maintaining tension is for the person to anticipate the tic (tension), prepare for the high-risk situation (tension), employ strategies to control the tic (tension), re-embark on an overactive style of action (tension). Treatment based on a CP model would thus necessarily focus on reduction of background sensory-motor activation by both cognitive and behavioral means. In the CP model, the immediate precursor of the tic onset is background tension and hyper-Vigilan to sensory state. Reducing tension overall however requires addressing input from four separate sources (see Fig. 6): muscle tension itself, overactive style of action, effortful preparation in anticipation of the high risk situation, and the maintaining coping behaviors (3C’s). The premonitory urge is a combination of sensory awareness, over-prepared muscle tension, attentional focus and the subsequent attribution of inevitability of the tic onset which might feed the attentional focus (see Fig. 3). Such heightened sensory awareness could be targeted and attenuated by producing habituation through interoceptive exposure to sensations coupled with response prevention. The response prevention would lead to eventual extinction both of the attentional focus behavior maintaining hypersensitive awareness and of the attribution according significance to the premonitory urge, and to the subsequent imminence of the tic (see Fig. 3). Cognitive strategies would also be useful to challenge hyper-attentiveness to self- and excessive self-consciousness and concern over self-image.

The onset of the tic itself is spurred on by the high degree of background muscle tension (Fig. 6), but in addition there seems a low degree of flexibility in the tic-affected muscle itself. The tic implicated muscle tends to be in an all or nothing state of tension (although never completely at zero tension level). Retraining the muscle to achieve more graded levels of tension changes its habitual use, and at the same time gives the person more flexibility and control over muscle use (O’Connor et al., 1995). There is also, in some tic-affected muscles, the tendency to involve more muscles than necessary in the action, for example blinking with cheek muscles in addition to eyelid muscles. Such unnecessary over-investment of effort in response can be normalized through education.

Such education in muscle use involves normalization of muscle use, exclusion of non-relevant muscles, use of appropriate effort and developing the ability to discriminate and articulate, with the help of EMG, the parts of a block response (ex. learning to blink with eyelid rather than cheek muscle). Another technique aimed at preventing tension is “prevention by relaxation”, which involves monitoring the tic-affected muscle to keep it in a state of relaxation in order to prevent the tic. This technique can be learnt and practiced as a form of habit reversal (O’Connor and Gareau, 1994, Pg 66). Rather than containing or suppressing the tics, if the person focusses on relaxation of the tic-implicated muscle, then the tic is prevented. A similar relaxation response is produced in 2 cases by Euers & Van De Wetering (1994). Competing slow response development has also been used as a treatment strategy by Miltenberger, Fuqua and Woods (1998).
However, the CP model predicts that prevention by relaxation is more likely to reduce muscle tension itself than another type of CR in the long run. The CR component, crucial to HR, is likely, by itself, to result in increasing tension as the muscle contracts antagonistically to the tic.

At the same time coping strategies and appraisals (see Fig. 5) maintaining muscle tension and stress, indeed all effortful ways of disguising or holding in the tic would ideally be extinguished and replaced by relaxation responses. Such coping strategies would include containment strategies which result in both enhanced vigilance to stress cues, and preoccupation with the tic which, in turn, can amplify tension and sensory awareness. The preferred strategies then to reduce tension involve: increasing muscle flexibility, decreasing behavioral activation, correcting the perfectionist beliefs, and the secondary appraisals and coping behaviors maintaining arousal, coupled with an education in economic muscle use.

The specific link in the CP model between perfectionism and behavioral activation could have implications for the treatment of hyperactivity in children, if we see overactivity in adults as a more mature form of childhood hyperactivity. The seemingly impulsive nature of hyperactivity may be driven rather by perfectionist beliefs about personal organization. Interestingly, stimulants like MPH reportedly give subjective feelings of confidence and this may explain their paradoxical effect on hyperactivity. The effect may be produced not via motor systems as such, but through alleviating perfectionist driven feelings of frustration by feelings of accomplishment. The desire in the child with TS + hyperactivity to switch amongst many tasks or for example, see every room in a building may not reflect a loss of inhibition but rather a perfectionist need to do as much as possible. In the same way, other disinhibited behaviors in more severe TS, such as “forbidden touching” or other risky behaviors, may be a consequence of an over-prepared reaction aimed to control, not to abandon, behavior. In other words, knowing that a behavior is forbidden would lead the normal person to have confidence in their ability to know not do it. In TS however the person immediately prepares actively to not do the action which, of course, paradoxically emphasizes the possibility that it could occur. This over-investment in restraining an activity maintains the importance of not doing it, and reinforces the thought of loss of control. This thought then leads to “tests” of control like half-extending and retracting the hand as though about to touch something forbidden. Such near-miss tests are intended to demonstrate control or rather disconfirm the idea that loss of control might occur. Another example of this over-investment in preventing an action is the case of a TS patient whose phonic tic was to bark like a dog. Paradoxically, it was the thought that he might bark like a dog and the strong desire not to do so that led him to bark as a test of his self-control over barking. Throat clearing urges in libraries, where noise is forbidden, are frequently preceded by preoccupations with controlling such urges. The over-investment and focus on the throat muscles in turn provoking the urge. This pattern of concern clearly resembles the obsessional person who constantly reminds himself not to forget, and who reacts to the thought of doing something “as if” it was equivalent to having done it.

An important issue is whether the CP treatment model outlined here and applied to TD with principally one dominant tic, can be generalized to more severe cases of TS. The classification of TS by vocal or phonic tic is clearly an historical accident and the CP model outlined here applies equally to phonic and motor tics alike. However, in severe ticcing where associated behavior is more self-destructive, there may be less place to unravel contributing sensory and cognitive factors. The preoccupation with ticcing may take on a life of its own and create a separate very short-term reinforcement schedule. A constant cycle of premonitory urge and frus-
tration will produce and, in turn, be maintained by increasingly disinhibited tension/sensation relieving tic behavior. The original perfectionism or hyper-sensitivity will appear remote from the increasingly local state dependent reinforcement of the tic cycle. Here clearly habituation to a sensory state and increasing tolerance of frustration might be essential initial elements to a treatment program prior to cognitive restructuring. There is indeed some evidence that exposure to a premonitory urge is more effective in severe TS than HR (Verdellen, Keijsers, Hoogduin, & Cath, 2002).

Further predictions of the current CP model are that: (1) targeting beliefs underlying perfectionist styles of action will improve attention/arousal and mood regulation in TD, TS, and HD; (2) addressing such cognitive factors will also aid general muscle relaxation and prevent tension build-up in tic-affected muscles; (3) addressing cognitive appraisals about tics will aid habituation to hypersensitivity by shifting attentional attribution and self-focus; (4) all tics and habits in TD/TS/HD will show a characteristic situation/activity profile associated with onset. (There is strong covariation between HD and TD, and it is likely that HD serves a similar function to TD but in response to more complex emotions.); (5) the muscles implicated in tics are likely to be linked by usage or habitual expression to an action–frustration cycle in high-risk tic onset situations/activities; (6) preventing tension build-up by addressing the four principal inputs to excessive activation (see Fig. 6) will be a more effective intervention in the long-run than habit reversal, and more likely to prevent tic substitution; (7) reducing the level of motor activation will result in changes in experimental motor performance as measured, for example, in studies of motor preparation, fine motor skills, and response inhibition; (8) modifying tic behavior will impact on dopamine and serotonin regulation; (9) individual differences in the premeditated cognitive/emotive activity profiles associated with tic habits may explain part of the variance in differential responses to types of medication; (10) the CP model will apply equally to TD and TS, although treatment might differentially target emotional and sensory or cognitive reinforcers of the tic behavior depending on tic severity.

References


