

TOURETTE SYNDROME: SCHOOL-BASED INTERVENTIONS FOR TICS AND ASSOCIATED CONDITIONS

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Tourette syndrome (TS) is a neurological disorder characterized by motor and phonic tics that follow a fluctuating pattern of severity, intensity and frequency. TS is often associated with other conditions such as attention-deficit/hyperactivity disorder, obsessive-compulsive disorder and learning difficulties. This complex phenotype affects the academic performance and social adjustment of pupils with TS. However, little data are available on the role of school-based interventions. This paper reviews the educational interventions for TS and consolidates evidence on the fundamental role of school in the management of the behavioral and learning difficulties that pupils with TS experience. The study aims to investigate the educational repercussions of tics and associated conditions, to examine the educational interventions suggested for their management and to highlight the magnitude of the role of teachers, peers and parents. Moreover, it provides future directions for the improvement of school-based interventions along with suggestions for further research.

Tourette syndrome (TS) is a neurological disorder that is characterized by motor and phonic tics that follow a fluctuating pattern of severity, intensity and frequency (Hansen, 1992; Leckman et al., 1998; Lin et al., 2002; Robertson et al., 1999). TS is often associated with other conditions such as attention-deficit/hyperactivity disorder (ADHD), obsessive-compulsive disorder (OCD) and learning difficulties. Additionally, antisocial behavior, inappropriate sexual activity, exhibitionism, aggressive and violent behavior, discipline problems, sleep disturbances and self-injurious behaviors are found in a remarkable percentage of individuals with TS (Robertson, 2000).

TS was considered to be a very rare disorder with an estimated prevalence rate of about 0.05% (5 per 10,000 individuals) (Bruun, 1984; Caine, et al., 1988). More recent studies have revealed that the prevalence is about one percent for children and adolescents from 5 to 18 years (Robertson, 2005). TS is much more common in males; the male: female ratio is 3:1 to 4:1, and it appears cross-culturally with the same clinical characteristics (Staley, Wand, & Shady, 1997).

Most researchers argue that TS is an inherited condition (Robertson, 2000). Imbalances in dopamine and serotonin have been reported, but it is still unknown how these two systems are involved in the pathogenesis of TS (Singer & Minzer, 2003). Anatomical and neuroimaging studies have provided further evidence for abnormal basal ganglia function in TS (Albin & Mink, 2006). It has also been speculated that perinatal factors, such as prematurity and streptococcal infections, predispose an individual to TS, but there is not strong research evidence to support these claims (Robertson, 2000).

Several diagnostic systems are used in TS, including the International Classification of Diseases and Health Related Problems (ICD-10) (World Health Organization, 1992), the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV) (American Psychiatric Association, 1994) and the Yale Global Tic Severity Scale (Leckman et al., 1989). A common feature of all these assessment tools is that diagnosis is mainly based on the identification of tics. Therefore, tics are universally considered as the clinical hallmark on which the diagnosis of TS is based.

However, there are still many cases that remain undiagnosed or misdiagnosed. The failure to identify repetitive sounds and movement as tics has been cited as a primary reason for the misdiagnosis of the syndrome (Bruun, 1984). This failure usually happens because patient symptoms are attributed to other conditions, such as hyperactivity, nervousness, habits, allergies, asthma, dermatitis (Jankovic, 2001), and to severe psychopathologic disorders, such as psychogenic tremor (Kulisevsky, Berthier, Avila, Ginorell, & Escartin, 1998).

Recent literature underlines that TS should not be considered only as a motor disorder, but emphasis should be given to the behavioral and cognitive profile of individuals with TS (Cavanna, Servo, Monaco, & Robertson, 2009). This complex phenotype causes children with TS to have numerous difficulties in school, which impact their academic performance and social adjustment. To date, several studies have investigated the impact of medical and behavioral treatments on tics and associated conditions of TS, but little data are available on the role of school-based interventions.

The purpose of this paper is to review the educational interventions for TS suggested in the literature and to consolidate evidence on the fundamental role of school in the management of the behavioral and learning difficulties that pupils with TS experience. This study aims to investigate the educational repercussions of tics and associated conditions and to examine the educational interventions suggested for their management. This paper also aims to highlight the magnitude of the role of teachers and peers and to underline the importance of parental involvement. Moreover, it provides future directions for the improvement of school-based interventions along with suggestions for further research.

Profile of pupils with TS: Tics and associated conditions

Tics are defined as *sudden, repetitive, stereotyped motor movements or phonic productions that involve discrete muscle groups* (Leckman, King, & Cohen, 1999, p.23), which *occur without any prolonged forethought by the person* (Kutscher, 2005, p. 147). Both motor and phonic tics are frequently preceded by premonitory sensations that cause discomfort and that are relieved after the execution of the tic (Jankovic, 2001).

Motor tics can be described as simple or complex. Simple motor tics are sudden, brief and meaningless movements, such as eye blinking, facial grimacing, mouth movements, head jerks, shoulder shrugs, and arm and leg jerks (Leckman et al., 1999). Complex motor tics are sudden, stereotyped movements of longer duration that seem more goal-directed and are very rarely occurring in the absence of simple motor tics. Examples of complex motor tics include grooming-like gestures, gyrating, bending, self-abusive acts, copropraxia and echopraxia (Leckman et al., 1999). The degree of impairment and disruption associated with particular motor tics varies depending on the number, frequency, intensity and complexity of specific tics, along with the impact of the tics on the individual's self-esteem, family life, social acceptance, school or job functioning and physical well-being (Leckman, 2003).

Phonic or vocal tics are tics that involve noises; similar to motor tics, they can also be grouped as simple or complex. Simple phonic tics are *fast, meaningless sounds or noises that can be characterized by their frequency, duration, volume, intensity and potential for disruptive speech* (Leckman et al., 1999, p.26). Simple phonic tics may be sniffing, throat clearing, grunting, barks and high-pitched squeaks. Complex phonic tics include more syllables, words, phrases and strange patterns of speech in which changes in terms of volume, rate and rhythm occur. Because of their distracting nature, phonic tics can cause significant troubles in school for pupils with TS, especially in peer relationships.

The course of tics is significant because symptom patterns do not only change in the short term but are likely to change over the individual's life (Robertson, 2000). From age 7 to 12 years, tics have the greatest effect on a child's self-esteem as well as on peer and family relationships (Leckman, Bloch, Scahill, & King, 2006). Leckman et al. (1998) found that in 36 individuals with TS, the mean tic onset was 5.6 years, and the onset was followed by a progressive pattern of worsening. The most severe tics were displayed at the average age of 10 years and caused significant problems for the children in school. The severe period of tics tends to be followed by a decline in tic severity (Leckman et al., 1998). The study of Bloch et al. (2006) supported these results, finding that over half to two-thirds of individuals with TS experience a reduction or complete resolution of tic symptoms during adolescence. Tics may also wax and wane because of other reasons, such as levels of stress (Cohen, Leckman, & Riddle, 1992).

There is a common agreement in the literature that TS is associated with ADHD, OCD and learning difficulties (Robertson, 2000). Other less common, associated conditions include self-injurious

behaviors, anxiety, personality disorder, depression, conduct disorder (Robertson, 2000) and autistic spectrum disorders (Canitano & Vivanti, 2007). It is possible that TS, OCD, ADHD and autism rest along a continuum with a similar underlying mechanism (Melillo & Leisman, 2004), and it is suggested that they are parts of a more complex disorder called the Developmental Basal Ganglia Syndrome (Palumbo, Maughan, & Kurlan, 1997).

Although tics are the main feature of TS, the presence of comorbidities, such as ADHD and OCD, are associated with more psychosocial and educational problems (Debes, Hjalgrim, & Skov, 2010). It is not uncommon for individuals with TS to report that they consider the comorbid conditions more distressing and disruptive than the actual tics themselves (Coffey & Park, 1997).

Medical and behavioral treatments

Pharmacological treatment is the most common treatment used for TS. Drugs that are usually used include the following: clonidine, guanfacine, haloperidol, primozide, risperidone, clozapine and tetrabenazine (Robertson, 2000) and aripiprazole (Davies, Stern, Agrawal, & Robertson, 2006). Drugs cannot eliminate tics entirely (Zinner, 2004). However, medication can contribute to tic attenuation, which will improve academic, psychosocial and family functioning (Zinner, 2004). Subsequently, pharmacological treatment can enhance the efficacy of non-medication based management (Zinner, 2004) by following and supporting the educational interventions and/or behavioral treatments (Carpenter, Leckman, Scahill, & McDougle, 1999). When medical treatment is indicated, the most prominent and problematic symptoms of the syndrome should be identified so that treatment takes place in the context of the single best-fit diagnostic category (Carpenter et al., 1999).

However, Robertson (2000) highlights that some of the drugs may have side effects, such as lack of motivation or lethargy, which can have consequences for the child's performance at school. Drawbacks associated with pharmacological treatments also include the possible lack of response to medication, difficulty with compliance and patient or parent reluctance (Sandor, 2003). Neurosurgical approaches have also been tried to reduce severe tics (Singer, 2005), and deep brain stimulation is gaining ground as a potential therapy for severe tics (Vandewalle, van der Linden, Groenewegen, Caemaert, 1999).

Behavioral treatments have been often implemented in TS as well; however, the outcomes have been contradictory (Carr & Chong, 2005; King, Scahill, Findley, & Cohen, 1999). Behavioral treatments used for TS include the following: massed negative practice, assertiveness training, self-monitoring, cognitive therapy and psychotherapy (King et al., 1999). However, such interventions were proven unsuccessful or counterproductive when aversive consequences were employed (Packer, 2005; Piacentini & Chang, 2001).

Habit reversal therapy (HRT) is the recent behavioral treatment of choice because it has been shown to be the most effective tic management method. HRT is based on the development of competing responses to tics and the enhancement of motivation (Piacentini & Chang, 2005). Clarke, Bray, Kehle, and Truscott (2001) investigated the effects of a two-component treatment package composed of habit reversal and self-modeling on the decrease of the tic frequency in 4 school-age students diagnosed with TS. Clarke et al (2001) found that three of the four students exhibited substantial decreases in their tics, which were maintained during a five- to ten-week follow up. Tics in the fourth student were reduced moderately. However, results should be treated carefully given the small number of participants in this study. Although several studies demonstrate that the use of HRT has shown promise in treating tics and associated impairments (Piacentini & Chang, 2005), it is less likely to be fruitful in young children, given that its success depends on awareness of premonitory sensory urges (Zinner, 2004). The literature also suggests that parent training can be effective when comorbid ADHD is present (Kazdin, 2003). Cognitive-behavioral treatments, such as exposure and response prevention, remain the first choice for the treatment of OCD, especially when there is significant anxiety or phobic avoidance (Pediatric OCD Treatment Study, 2004).

School-based interventions for TS

It is estimated that the prevalence of TS in the mainstream school population ranges from 0.56% to 3% (Hornsey, Banerjee, Zeitlin, & Robertson, 2001; Kurlan, Whitmore, Irvine, McDermott, & Como, 1994; Mason, Banerjee, Eapen, Zeitlin, & Robertson, 1998; Wang & Kuo 2003). Research has shown that the prevalence of TS is higher in special education settings. Comings, Hines, and Comings (1990) screened 3034 pupils referred for psychoeducational assessment in a southern Californian school district in the United States and estimated that 12% of all children in special education classes had TS.

The previous findings show that a remarkable proportion of children with TS are being educated both in mainstream and special educational settings. This prevalence renders necessary the broad implementation of school-based interventions for TS and associated conditions. The need for school-based interventions is further justified by the fact that, as we have seen above, the onset of tic symptoms coincides in terms of time with the onset of school life. These symptoms reach a peak at the age of about ten years.

Children with TS are five times more likely than their peers to need special education services (Kurlan et al., 1994). Debes et al. (2010) found that in a population of 314 children with TS, 59.0% had some kind of educational problems that needed support or special education. Despite the fact that there are several similarities among individuals with TS, it is of crucial importance to acknowledge that each individual presents a unique clinical and educational profile.

Carter et al. (1999) argue that although not all children with TS have special educational needs, specific educational interventions are necessary when (a) a pupil is falling behind academically; (b) tics are so frequent and forceful that they interfere with the child's learning or participation in the classroom; (c) a pupil has important difficulties in peer relationships; (d) the pupils' self esteem is in danger. Other indicators for the necessity of special education could include feelings of frustration, a negative change in attitude regarding school or teachers or behavioral problems that interfere with the school life in any manner (Packer, 2005). In any of these cases, a differential approach should be adopted. The unique needs of each individual pupil with TS should be considered and included when planning and implementing any educational assessments and interventions. To that end, a number of strategies involving teachers, peers and families could be applied.

Teacher attitude is critical for the school experience for children with TS. Grace and Russell (2005) conducted a qualitative study of 26 children with TS and found that the children who were happiest and most successful were those who felt that teachers were understanding and respectful of their feelings and needs. Therefore, teachers should be very well informed about the nature of the syndrome and its possible educational implications. However, several studies show that teachers do not have a good understanding of what exactly TS is (Chowdhury & Christie, 2002; Wilson & Shrimpton, 2002), and it is very likely that they do not have any relevant training (Packer, 1997).

The organization of training days for teachers can be very beneficial to enhance teachers' knowledge about the syndrome and its related special educational needs (Chowdhury & Christie, 2002). Additional provisions may include ongoing collaborative consultation models between the school psychologist and the teachers (Kepley & Connors, 2007). Communication with parents can further facilitate the understanding of the educational and behavioral profile of a child with TS (Grace & Russell, 2005). Collaborative models between the school and the parents, such as a daily report card system, can further enhance academic planning and improve behavioral strategies (Kepley & Connors, 2007). However, the involvement of parents in the education of their children can be very tiring and time consuming, so school psychologists have an important role to play in relieving parents of such a burden (Grace & Russell, 2005).

Teachers should also keep in mind that symptoms tend to appear less problematic when the pupil with TS is engaged in school activities at a pace that does not add increased pressure (Kepley & Connors, 2007). Teachers can provide extended time limits, give the option of alternative ways to produce work, allow pupils to choose their position in the classroom, reduce homework (Packer, 1997, 2005) and allow pupils to make short scheduled breaks (Kepley & Connors, 2007). Other classroom accommodations may include untimed tests and test taking in private rooms (Zinner, 2004). Motor tasks, including handwriting, can be affected by frequent tics, so scribes or tape recorders may be useful for children with functional writing difficulties (Zinner, 2004). Given the heterogeneity of the population of pupils with TS, these practical implications might not have equally beneficial outcomes for all pupils. Hence, teacher flexibility and continuous assessment are of crucial importance.

Another preventive strategy that is gaining ground in tic management in the classroom is altering antecedent stimuli (Watson, Dufrene, Weaver, Butler, & Meeks, 2005). Tics are sensitive to specific environmental incentives (Watson & Sterling, 1998; Woods, 2001). Watson et al. (2005) used functional assessment to find out which stimuli may trigger tics in school settings. They found that once these incentives were defined, they could be avoided or altered.

When tics appear in the classroom, their management is of major importance because the response of teacher and classmates can make a significant difference in the child's school performance. Expressing frustration and anger does not help the pupil with TS to stop performing the tics. Negative reactions can make pupils with TS feel more anxious, isolated, frustrated or hostile, and they may even cause the worsening of tics (Carter et al., 1999). Whenever possible, teachers should ignore the performance of tics. Alternatively, teachers can give permission to the pupils with TS to leave the classroom for a short time to release the tics that they have been suppressing (Packer, 2005). Since the symptoms of the syndrome may fluctuate, teachers must reevaluate and modify strategies according to the unique needs of each pupil at a particular period of time.

Schools can also take action to facilitate the social adjustment of pupils with TS. Children with TS often experience social rejection and isolation (Friedrich, Morgan, & Devine, 1996). They are usually less popular, at a higher risk for having poor peer relationships, and are more often targets of peer teasing than their classmates (Debes et al. 2010; Shady, Fulton, & Champion, 1988). Rosen (1996) provides a personal account and reports that he was often mocked and mimicked by other children.

If classmates are informed about the syndrome, they can be more tolerant and supportive as well as ignore the possible disturbance caused by tics (Carter et al., 1999). Peer education may contribute to the reduction of teasing, bullying, avoidance and tearful reactions by classmates (Kepley & Conners, 2007). In a broader context, peer tutoring interventions have been suggested as a way of increasing appropriate social behavior in the classroom (Bolich, 2001). Circle of friends is a strategy that can be used to improve self-esteem and relationships among classmates (Chowdhury & Christie, 2002). Murphy and Heiman (2007) found that group work could help children with TS to understand and deal with their condition. Such groups also provide children great opportunities for social interactions with peers and give parents the chance to meet and share their understanding of their children's needs. Collaboration among the parents of children with TS could be very valuable for effective evaluation and treatment (Carter et al., 1999). The school psychologist could also be involved and facilitate these meetings.

The self-awareness of pupils with TS about their condition can enhance their school performance as well. Lambert and Christie (1998) provided an example of social skills training for children with TS. They formed a social skills group for boys with TS to help them build their confidence, raise their self-esteem, improve their communication and interpersonal skills, increase self-awareness and awareness of others and reduce anxiety and stress related to social interaction. By the end of the group, there was an improvement in the self-esteem of the participants, and some of the children were so involved in the project that they even managed to stop their tics (Lambert & Christie, 1998).

Supplementary school-based interventions for associated conditions

ADHD is the most commonly reported disorder that occurs in children with TS, with prevalence's as high as 60% to 70% (Coffey et al., 2000; Spencer et al., 1998). The precise relationship between TS and ADHD has not yet been clarified and is still a matter of debate in the literature (Rizzo et al., 2007). However, it seems that patients with TS and comorbid ADHD have worse a psychosocial and cognitive profile in comparison to patients with TS (Brand et al., 2002). ADHD in the setting of TS is intimately associated with academic difficulties, peer rejection and family conflict (Carter et al., 2000; Hoekstra et al., 2004; Peterson, Pine, Cohen, & Cook, 2001; Spencer et al., 2001; Sukhodolsky et al., 2003). Thus, the presence of comorbid ADHD should be adequately assessed on time and treated appropriately (Rizzo et al., 2007).

ADHD in TS is influenced by distraction from the tics themselves and by attempts to inhibit the tics (Cavanna et al., 2009). Pupils with TS and ADHD will possibly show inattention, poor concentration and may be very destructive (Carroll & Robertson, 2000). Concentration difficulties can interfere seriously with learning (Debes et al., 2010) and cause many school problems (Abwender et al., 1996; Dykens et al., 1990; Schuerholz, Baumgardner, Singer, Reiss, & Denckla, 1996). Additionally, pupils with TS and ADHD may have difficulties converting words into concepts and following instructions. They may also deviate from class activities.

Carter et al. (1999) argue that educational interventions for children who have TS and ADHD should be similar to those for children who have ADHD alone. Such educational interventions include behavioral strategies in the classroom that improve attention, reduce impulsive responding and improve organizational skills. Examples of behavioral strategies constitute the use of token economies and of daily report card systems (e.g. Morisoli & McLaughlin, 2004). Lessons should be highly structured and

include well-defined expectations and clear consistency. Teachers should reward adaptive and positive behaviors and at the same time ignore disruptive behaviors (Carter et al., 1999). Moreover, educational interventions should consider the context in which the behavioral problem takes place so that the educational environment is clear of any possible sources of distraction (Zinner, 2004). Other researchers highlight the effectiveness of the combination of both stimulant medication that regulates the neural substrate of behavioral inhibition and the respective executive functions and behavioral interventions implemented in classroom (Morisoli & McLaughlin, 2004).

Pupils who have TS along with ADHD face more difficulties in social adaptation than children with TS without ADHD (Carter et al., 2000). Existing studies pinpoint peer relationship problems of children with ADHD and consider multi-component interventions highly beneficial to ameliorate the social problems of children with the disorder (Ozdemir, 2010). Therefore, there is an even greater need for social skills training that aims to improve social competences with peers and adults. Teachers can support and motivate pupils with TS and ADHD to do things that improve their relationships with peers. For instance, teachers can show to pupils how to become more patient, share things, let their classmates speak first and keep quieter voice levels during the lesson (Carroll & Robertson, 2000).

Another commonly associated condition is OCD. Recent literature shows that OCD and TS are strongly associated but that the estimated percentage of comorbidity varies from 11% to 80% (Robertson, 2000). OCD is far more common in children and adults with TS than without TS (American Psychiatric Association, 2000). A substantial number of clinical series have found that individuals with a tic-related form of OCD are more likely to report obsessions of symmetry and exactness. They are also likely to feel an urge to do and redo activities to achieve a sense of completion or a sense of things looking, feeling, or sounding *just right* (Kwak, 2003; Woods et al., 2005). OCD can interfere with learning because it causes marked distress and is time-consuming (American Psychiatric Association, 1994). Children with OCD may be distracted by intrusive thoughts and the desire to perform compulsive behaviors (Scahill, Ort, & Hardin 1993). The anxiety caused by intrusive thoughts or the need to perform a compulsive behavior is pressing and difficult to resist (Carter et al., 1999).

Children with TS can become very anxious if they realize that their symptoms are obvious to others. Therefore, teachers who suspect symptoms of OCD should avoid direct confrontation with the child as this can lead to anxiety and embarrassment (Carter et al., 1999). A variety of modifications can be made for children who have TS along with OCD, including help with note taking and leeway or limits relevant to activities in which children with OCD tend to perform symptoms (Carter et al., 1999). Other strategies that can be helpful include the option of completing the work orally, short breaks, opportunities to switch tasks and continuous encouragement to keep on working despite the presence of OCD symptoms (Carter et al., 1999). Pupils with OCD tend to have particular difficulties with transitions from one task to another because they are unable to understand that the task has finished. In such cases teachers should allow pupils to go back to the task at a later time.

Furthermore, it has been estimated that at least half of the children with TS have learning difficulties which often are related to mathematical and language perception and comprehension difficulties (Brookshire, Butler, Ewing-Cobbs, & Fletcher, 1994; Comings, 1990). These specific learning difficulties are frequently caused by neuropsychological deficits in the domains of visual-motor integration and executive functioning (Schultz, Carter, Scahill, & Leckman, 1999).

Hence, management of writing tasks and tasks that require organization skills can be very difficult and time-consuming for pupils with TS. Silver (1988) found that writing was slow and preservative and that children with TS needed more time than their peers to complete writing tasks. Teachers should focus on the quality rather than on the quantity of the written work produced. When significant difficulties in writing are present, written tasks may be given without time restrictions.

Discussion

In the present paper, we reviewed the available literature related to the role of school in coping with TS symptoms and associated conditions. As shown above, the prevalence of TS, both in the mainstream and special sector, places a premium on the implementation of school-based interventions. In light of the significant position that school occupies in every pupil's life, teachers can and should play a paramount, multidimensional role, not only in pupils' academic progress but also in their social competence because these two aspects of functioning are interdependent.

For instance, teachers can play a role in diagnosis. Given that misdiagnosis and under diagnosis preclude several individuals with TS from appropriate educational interventions and suitable medical and behavioral treatment; teachers should contribute to the diagnosis of TS in the school population. The accomplishment of such a task requires the necessary knowledge, level of awareness and consequent understanding of the syndrome by the teachers. Research reveals the lack of information about TS. In particular, teachers working in mainstream schools probably need higher levels of support than their colleagues working in special settings because they are less familiar with TS. Therefore, the enhancement of teacher knowledge about the syndrome would further help them to handle more effectively behavioral and learning difficulties associated with TS.

Despite the fact that several publications provide suggestions for accommodations and modifications that target the construction of the most helpful classroom environment, the complexity of the TS phenotype should be taken into account in the implementation of every intervention. Accordingly, teachers should be flexible regarding the strategies they use; these strategies should be tailored to every pupil with TS, regularly evaluated and modified depending on the course of TS symptoms and associated difficulties. Moreover, teachers should play a proactive role in peer education by working on projects aimed at the acceptance of differences, and, in particular, the understanding and tolerance towards challenging behaviors of pupils with TS by their classmates.

Apart from the implementation of suitable educational strategies and special modifications in the classroom, teachers should also work in close collaboration with the parents of children with TS. Mounting evidence suggests that a mutually enriching relationship between school and family optimizes the educational outcome. Such collaboration would also relieve families of pupils with TS who experience negative feelings and loneliness. The involvement of professionals in extracurricular activities and support services, and the subsequent adoption of an interdisciplinary model of collaboration, should be an organic part of this procedure.

However, teachers should not carry the weight alone. A possible solution could involve the teaching assistants and school psychologist but again the issue of awareness on the particular needs of pupils with TS would occur. Collaboration with outreach services with expertise in TS would facilitate schools in the organization of in-service training. However, TS awareness should not be limited to schools. The consideration of TS into an ecological framework renders necessary the enhancement of public awareness. School should not only provide adequate educational opportunities but also play a dynamic role at the community level.

Overall, the study of a complex phenotype such as TS reveals the necessity of the convergence among medical, psychological and educational sciences as well as among the professionals who serve them, to unlock the potential of students with TS and to ensure the best possible functioning in the academic, psychosocial and family domain.

Future research

Our review brings into focus a number of potentially fruitful topics for future research. There is little available research on the effectiveness of teachers' strategies both for the prevention and management of tics. Several limitations of the investigations were identified concerning methodological issues such as the small number of participants and the limited sources of data, which usually involve parents of pupils with TS and exclude personal accounts.

Case studies involving qualitative methods, such as interviews, would contribute to a better understanding of the syndrome and subsequent improvement of educational provisions. Comparative studies evaluating the efficacy of teaching strategies and behavioral modifications would also give useful insights. Moreover, longitudinal and follow-up studies would verify the maintenance of long-term gains after the implementation of different approaches and evaluate their possible impact on the course of tics.

Interventions that aim toward behavior modification without emphasizing the psychopathology of the syndrome could be very beneficial for children with TS. Positive Behavior Support aims to change an individual's life through a child-centered approach that takes into account the perspectives of teachers, peers and parents. Researchers and practitioners could apply this approach with children who have TS to evaluate the potential benefits of such an approach both on their social adjustment and academic performance.

Children with TS face significant difficulties in their social adjustment, often suffer from social embarrassment and have low self-esteem, which affects their quality of life. Future research should shed light on the personal networks of pupils with TS as well as on the types and the levels of social support that they receive from their social networks.

Pupils with TS and ADHD are usually instructed as if they have ADHD alone. It is a matter of future research to investigate whether this approach is the most appropriate. The same subgroup of pupils with TS is at greater risk of facing problems with their social adjustment. Therefore, it is generally accepted that there is a need for social skills enhancement. Several strategies are suggested for social skills training, such as instruction, modeling and role-playing. A future challenge is to find out which approach is the most appropriate to meet the needs of pupils with TS and to examine the impact of age-related factors to the success of the interventions.

Moreover, the specific modifications and strategies used for the pupils who have OCD in the setting of TS should be evaluated on the basis of the fluctuating course that the symptoms of these two conditions have. Another topic that needs to be clarified concerns the relation between autistic spectrum disorders and TS. Although a number of studies indicate that TS and autistic spectrum disorders may share a common denominator, much remains to be determined concerning the educational implications of such comorbidity. Therefore, greater research efforts in the future are welcomed.

References

- Abwender, D., Como, P., Kurlan, R., Parry, K., Fett, K., Cui, L., Plumb, S., & Deeley, C. (1996). School problems in Tourette's Syndrome. *Archives of Neurology*, *53*, 509-511.
- Albin, R. L. & Mink, J. W. (2006). Recent advances in Tourette syndrome research. *Trends in Neurosciences*, *29*, 175-182.
- American Psychiatric Association (2000). *Diagnostic and Statistical Manual of Mental Disorders*, 4th edition, text revision (DSM-IV-TR). Washington, DC: American Psychiatric Association.
- Bolich, B. J. (2001). Peer tutoring and social behaviors: A review. *International Journal of Special Education*, *16*, 16-30.
- Bloch, M. H., Peterson, B. S., Scahill, L., Otko, J., Katsovich, L., Zhang, H., & Leckman, J.F (2006). Adulthood outcome of tic and obsessive-compulsive symptom severity in children with Tourette syndrome. *Archives of Pediatrics and Adolescent Medicine*, *160*, 65-69.
- Brand, N., Geenen, R., Oudenhoven, M., Lindenborn, B. van der Ree, A., Cohen-Kettenis, P., & Buitelaar, J. K. (2002). *Brief report: Cognitive functioning in children with Tourette's syndrome with and without comorbid ADHD*. *Journal of Pediatric Psychology*, *27*, 203-208.
- Brookshire, B., Butler, I., Ewing-Cobbs, L., & Fletcher, J. (1994). Neuropsychological characteristics of children with Tourette Syndrome: Evidence for a nonverbal learning disability? *Journal of Clinical and Experimental Neuropsychology*, *16*, 289-302.
- Bruun, R. D. (1984). Gilles de la Tourette's syndrome: an overview of clinical experience. *Journal of the American Academy of Child and Adolescent Psychiatry*, *23*, 621-624.
- Caine, E. D., McBride, M. C., Chiverton, P., Bamford, K. A., Redliess, S., & Shiao, J. (1988). Tourette's syndrome in Monroe county school children. *Neurology*, *38*, 472-475.
- Canitano, R., & Vivanti, G. (2007). Tics and Tourette syndrome in autistic spectrum disorders. *Autism*, *11*, 19-28.
- Carpenter, L. L., Leckman, J. F., Scahill, L., & McDougle, C. J. (1999). Pharmacological and other somatic approaches to treatment. In Leckman, J. F., & Cohen, D. J. (Eds.) *Tourette's syndrome – Tics, obsessions, compulsions: Developmental psychopathology and clinical care* (pp. 370-398). New York: John Wiley and Sons Inc.
- Carr, J. E., & Chong, I. M. (2005). Habit reversal treatment of tic disorders: A methodological critique of the literature. *Behavior Modification*, *29*, 858-875.
- Carroll, A., & Robertson, M. (2000). *Tourette syndrome: A practical guide for teachers, parents and carers*. London: David Fulton.
- Carter, A. S., Fredine, N. J., Findley, D., Scahill, L., Zimmerman, L., & Sparrow, S. S. (1999). *Recommendations for teachers*. Hoboken, NJ: John Wiley & Sons Inc.
- Carter, A. S., O'Donnell, D. A., Scahill, L., Schultz, R.T, Leckman, J. F., & Pauls, D. L. (2000). Social and emotional adjustment in children affected with Gilles de la Tourette's syndrome: associations with ADHD and family functioning. *Journal of Child Psychology and Psychiatry*, *41*, 215-223.
- Cavanna, A. E., Servo, S., Monaco, F., & Robertson, M. M. (2009). The behavioral spectrum of Gilles de la Tourette Syndrome. *The Journal of Neuropsychiatry and Clinical Neurosciences*, *21*, 13-23.

- Chowdhury, U., & Christie, D. (2002). Tourette Syndrome: A training day for teachers. *British Journal of Special Education*, 29, 123-127.
- Clarke, M.A., Bray, M.A., Kehle, T.J., & Truscott, S.D. (2001) A school-based intervention designed to reduce the frequency of tics in children with Tourette's syndrome. *School Psychology Review*, 30, 11-22.
- Coffey, B. J., Biederman, J., Smoller, J. W., Geller, D. A., Sarin, P., Schwartz, S., & Kim, G. S. (2000). Anxiety disorders and tic severity in juveniles with Tourette's disorder. *Journal of the American Academy of Child and Adolescent Psychiatry*, 39, 562-568.
- Coffey, B., & Park, K. (1997). Behavioural and emotional aspects of Tourette Syndrome. *Neurologic Clinics*, 15, 277-289.
- Cohen, D., Leckman, J., & Riddle, M. (1992). Tourette's syndrome and tic disorders. In J. Noshpitz (Ed.), *Basic handbook of child psychiatry*. New York: Basic Books.
- Comings, D. (1990). *Tourette Syndrome and human behaviour*. Duarte, CA: Hope Press.
- Comings, D. E., Hines, J. A., & Comings, B. G. (1990). An epidemiologic study of Tourette syndrome in a single school district. *Journal of Clinical Psychiatry*, 51, 563-569.
- Davies, L., Stern, J. S., Agrawal, N., & Robertson, M. M. (2006). A case series of patients with Tourette's syndrome in the United Kingdom treated with aripiprazole. *Human Psychopharmacology: Clinical and Experimental*, 21, 447-453.
- Debes, N., Hjalgrim, H., & Skov, L. (2010). The presence of attention-deficit hyperactivity disorder (ADHD) and obsessive-compulsive disorder worsen psychosocial and educational problems in Tourette syndrome. *Journal of Child Neurology*, 25, 171-181.
- Dykens, E., Leckman, J., Riddle, M., Hardin, M., Schwartz, S., & Cohen, D. (1990). Intellectual, academic, and adaptive functioning of Tourette Syndrome children with and without attention deficit disorder. *Journal of Abnormal Child Psychology*, 18, 607-615.
- Friedrich, S., Morgan, S., & Devine, C. (1996). Children's attitudes and behavioural intentions toward a peer with Tourette Syndrome. *Journal of Pediatric Psychology*, 27, 307-319.
- Grace, R., & Russell, C. (2005). *Tourette's syndrome and the school experience: A qualitative study of children's and parents' perspectives*. *Australasian Journal of Special Education*, 29, 40-59.
- Hansen, C. (1992). What is Tourette Syndrome? In T. Hearle (Ed.), *Children with Tourette Syndrome: A parents' guide* (pp. 1-25). Rockville, MD: Woodbine House.
- Hoekstra, P. J., Anderson, G. M., Limburg, P. C., Korf, J., Kallenberg, C. G., & Minderaa, R. B. (2004). Neurobiology and neuroimmunology of Tourette's syndrome: an update. *Cellular and Molecular Life Sciences*, 61, 886-898.
- Hornsey, H., Banerjee, S., Zeitlin, H., & Robertson, M. (2001). The prevalence of Tourette syndrome in 13-14 year olds in mainstream schools. *Journal of Child Psychology and Psychiatry*, 42, 1035-1039.
- Jankovic, J. (2001). Tourette's syndrome. *The New England Journal of Medicine*, 345, 1184-1192.
- Kazdin, A. E. (2003). Problem-solving skills training and parent management training for conduct disorder. In A. E. Kazdin & J. R. Weisz (Eds.) *Evidence-based psychotherapies for children and adolescents* (pp. 241-262). New York: Guilford.
- Kepley, H. O., & Connors, S. (2007) Management of learning and school difficulties in children with Tourette syndrome. In D. W. Woods, J. C. Piacentini & J. T. Walkup, (Eds), *Treating Tourette syndrome and tic disorders: a guide for practitioners* (pp. 242-264). New York: Guilford Press.
- King, R. A., Scahill, L., Findley, D., & Cohen, D. J. (1999). Psychosocial and behavioral treatments. In J. F. Leckman & D. J. Cohen (Eds.), *Tourette's syndrome – Tics, obsessions, compulsions: Developmental psychopathology and clinical care* (pp. 43-62). New York: John Wiley and Sons Inc.
- Kulisevsky, J., Berthier, M. L., Avila, A., Ginorell, A., & Escartin, A. E. (1998). Unrecognized Tourette syndrome in adult patients referred for psychogenic tremor. *Archives of Neurology*, 55, 409-414.
- Kurlan, R., Whitmore, D., Irvine, C., McDermott, M. P., & Como, P. G. (1994). Tourette's syndrome in a special education population: a pilot study involving a single school district. *Neurology*, 44, 699-702.
- Kutscher, M. L. (2005). *Kids in the syndrome mix of ADHD, LD, Asperger's, Tourette's, bipolar and more!* London: Jessica Kingsley Publishers.
- Kwak, C., Vuong, K. D., & Jankovic, J. (2003). Premonitory sensory phenomenon in Tourette's syndrome. *Movement Disorders*, 18, 1530-1533.
- Lambert, S., & Christie, D. (2000). Social skills group for boys with Tourette's syndrome. *Clinical Child Psychology and Psychiatry*, 3, 267-277.
- Leckman, J. F., Bloch, M. H., King, R. A., & Scahill, L. (2006). Phenomenology of tics and natural history of tic disorders. *Advances in Neurology*, 99, 1-16.
- Leckman, J. F., Bloch, M. H., Scahill, L., & King, R. A. (2006). Tourette syndrome: The self under siege. *Journal of Child Neurology*, 21, 642-649.

- Leckman, J. F., King, R. A., & Cohen, D. J. (1999). Tics and tic disorders. In J.F. Leckman & D.J. Cohen (Eds.) *Tourette's syndrome - Tics, Obsessions and Compulsions: developmental psychopathology and clinical care*. New York: John Wiley and Sons Inc.
- Leckman, J. F., Riddle, M. A., Hardin, M. T., Ort, S. A., Swartz, K. L., Stevenson, J., & Cohen, J. (1989). The Yale global tic severity scale. *Journal of the American Academy of Child and Adolescent Psychiatry, 28*, 566-573.
- Leckman, J. F., Zhang, H., Vitale, A., Lahnin, F., Lynch, K., Bondi, C., Kim, Y. S., & Peterson, B. S. (1998). Course of tic severity in Tourette syndrome: The first two decades. *Pediatrics, 102*, 14-19.
- Lin, H., Yeh, C. B., Peterson, B. S., Scahill, L., Grantz, H., Findley, D. B., Katsoch, L., & Leckman, J. F. (2002). Assessment of symptom exacerbations in a longitudinal study of children with Tourette's syndrome or obsessive-compulsive disorder. *Journal of the American Academy of Child and Adolescent Psychiatry, 41*, 1070-1077.
- Mason, A., Banerjee, S., Eapen, V., Zeitlin, H., & Robertson, M. M. (1998). Prevalence of Tourette syndrome in a mainstream school population. *Developmental Medicine and Child Neurology, 40*, 292-296.
- Melillo, R., & Leisman, G. (2004). *Neurobehavioral disorders of childhood: An evolutionary perspective*. New York: Kluwer Academic/Plenum Publishers.
- Morisoli, K & McLaughlin, T. S. (2004). Medication and school interventions for elementary students with attention deficit hyperactivity disorder. *International Journal of Special Education, 19*, 97-106.
- Murphy, T., & Heiman, I. (2007). Group work in young people with Tourette syndrome. *Child and Adolescent Mental Health, 12*, 46-48.
- Ozdemir, S. (2010). Peer relationship problems of children with AD/HD: Contributing factors and implications for practice. *International Journal of Special Education, 25*, 184-193.
- Packer, L. E. (1997). Social and educational resources for patients with Tourette syndrome. *Neurologic Clinics, 15*, 457-473.
- Packer, L. E. (2005). *Tic-related school problems: Impact on functioning, accommodations, and interventions*. *Behavior Modification, 29*, 876-899.
- Palumbo, D. Maughan, A., & Kurlan, R. (1997). Tourette syndrome is only one of several causes of a developmental basal ganglia syndrome. *Archives of Neurology, 54*, 475-483.
- Pediatric OCD Treatment Study (POTS) Team (2004). Cognitive-behavior therapy, sertraline, and their combination for children and adolescents with obsessive-compulsive disorder: the Pediatric OCD Treatment Study (POTS) randomized controlled trial. *Journal of the American Medical Association, 292*, 1969-1976.
- Peterson, B. S., Pine, D. S., Cohen, P., & Cook, J. (2001). Prospective, longitudinal study of tic, obsessive-compulsive, and attention-deficit/hyperactivity disorders in an epidemiological sample. *Journal of the American Academy of Child and Adolescent Psychiatry, 40*, 685-695.
- Piacentini, J., & Chang, S. (2001). *Behavioral treatments for Tourette syndrome: State of the art*. In D. J. Cohen, J. Jankovic, & C. Goetz (Eds.), *Advances in neurology: Tourette syndrome (Vol. 85, pp. 319-332)*. Philadelphia: Lippincott Williams & Wilkins.
- Piacentini, J., & Chang, S. (2005). *Habit reversal training for tic disorders in children and adolescents*. *Behavior Modification, 29*, 803-822.
- Piacentini, J., & Chang, S. W. (2006). Behavioral treatments for tic suppression: Habit reversal training. *Advances in Neurology, 99*, 227-233.
- Rizzo, R., Curatolo, P., Gulisano, M., Virzi, M., Arpino, C., & Robertson, M. M. (2007). Disentangling the effects of Tourette syndrome and attention deficit hyperactivity disorder on cognitive and behavioral phenotypes. *Brain & Development, 29*, 413-420.
- Robertson, M. M. (2005). Tourette's syndrome, *Psychiatry, 4*, 92-97.
- Robertson, M. M., Banerjee, S., Kurlan, R., Cohen, D. J., Leckman, J. F., McMahon, W., Pauls, D. L., & Wetering, B. J. M. (1999). The Tourette syndrome diagnostic confidence index: Development and clinical associations. *Neurology, 53*, 2108-2112.
- Robertson, M.M. (2000). Tourette syndrome, associated conditions and the complexities of treatment. *Brain, 123*, 425-462.
- Rosen, A. (1996). Tourette's Syndrome; The school experience. *Clinical Pediatrics, 35*, 467-469.
- Sandor, P. (2003). Pharmacological management of tics in patients with TS. *Journal of Psychosomatic Research, 55*, 41-48.
- Scahill, L., Ort, S. I., & Hardin, M. T. (1993). *Tourette's Syndrome, Part I: Definition and diagnosis*. *Archives of Psychiatric Nursing, 7*, 203-208.
- Schuerholz, L., Baumgardner, T., Singer, H., Reiss, A., & Denckla, M. (1996). Neuropsychological status of children with Tourette's syndrome with and without attention deficit hyperactivity disorder. *Neurology, 46*, 958-965.

- Schultz, R. T., Carter, A. S., Scahill, L., & Leckman, J. F. (1999). Neuropsychological findings. In Leckman, J. F. and Cohen, D. J. (Eds) *Tourette's syndrome – Tics, obsessions, compulsions: developmental psychopathology and clinical care* (pp. 80-103). New York: John Wiley and Sons Inc.
- Shady, G. A., Fulton, W. A., & Champion, L. M. (1988). Tourette syndrome and educational problems in Canada. *Neuroscience and behavioural reviews*, 12, 263-265.
- Silver, A. (1988). Intrapsychic processes and adjustment in Tourette's Syndrome. In D. J. Cohen, R. D. Bruun & J. F. Leckman (Eds.), *Tourette Syndrome and tic disorders: Clinical understanding and treatment* (pp. 197-206). New York: John Wiley and Sons.
- Singer, H. S. (2005). Tourette's syndrome: From behaviour to biology. *The Lancet Neurology*, 4, 149-159.
- Singer, H. S., & Minzer, K. (2003). Neurobiology of Tourette's syndrome: Concepts of neuroanatomic localization and neurochemical abnormalities. *Brain Development*, 25, 70-74.
- Spencer, T., Biederman, J., Coffey, B., Geller, D., Faraone, S., & Wilens, T. (2001). Tourette disorder and ADHD. *Advances in Neurology*, 85, 57-87.
- Spencer, T., Biederman, J., Harding, M., O'Donnell, D., Wilens, T., Faraone, S., Coffey, B., & Geller, D. (1998). Disentangling the overlap between Tourette's disorder and ADHD. *Journal of Child Psychology Psychiatry*, 39, 1037-1044.
- Staley, D., Wand, R., & Shady, G. (1997). Tourette disorder: A cross-cultural overview. *Comprehensive Psychiatry*, 38, 6-16.
- Sukhodolsky, D. G., Scahill, L., Zhang, H., Peterson, B. S., King, R. A., Lombroso, P. J., Katsovich, L., Findley, D., & Leckman J. F. (2003). Disruptive behavior in children with Tourette's syndrome: Association with ADHD comorbidity, tic severity, and functional impairment. *Journal of the American Academy of Child and Adolescent Psychiatry*, 42, 98-105.
- Vandewalle, V., van der Linden C, Groenewegen, H. J., Caemaert, J. (1999). Stereotactic treatment of Gilles de la Tourette syndrome by high frequency stimulation of thalamus. *Lancet*, 353, 724.
- Wang, H.S., & Kuo, M.F. (2003). Tourette's syndrome in Taiwan: an epidemiological study of tic disorders in an elementary school at Taipei County. *Brain & Development*, 25, 29-31.
- Watson, T. S., & Sterling, H. E. (1998). Brief functional analysis and treatment of a vocal tic. *Journal of Applied Behavior Analysis*, 31, 471-474.
- Watson, T. S., Dufrene, B., Weaver, A., Butler, T., & Meeks, C. (2005). Brief antecedent assessment and treatment of tics in the general education classroom: a preliminary investigation. *Behavior Modification*, 29, 839-857.
- Wilson, J., & Shrimpton, B. (2002). What's normal? A reflection on Tourette syndrome. *Primary Educator*, 8, 6-8.
- Woods, D. W., Piacentini, J., Himle, M. B., & Chang, S. (2005). Premonitory Urge for Tics Scale (PUTS): Initial psychometric results and examination of the premonitory urge phenomenon in youths with tic disorders. *Journal of Developmental & Behavioral Pediatrics*, 26, 397-403.
- Woods, D.W. (2001). Habit reversal treatment manual for tic disorders. In D.W.Woods, & R. G. Miltenberger (Eds.), *Tic disorders, trichotillomania, and other repetitive behavior disorders: Behavioral approaches to analysis and treatment* (pp. 97-132). New York: Kluwer.
- World Health Organization (1992). *International Classification of Diseases* (10th edition). Geneva: World Health Organization.
- Zinner, S. H. (2004). Tourette syndrome-much more than tics, Part 2: Management tailored to the entire patient. *Contemporary Pediatrics*, 21, 38-49.