

Possible Role of Repetitive Practice of Activities Requiring Reflexive Responses in the Treatment of Tourette's Disorder

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We report 2 boys, 11 and 7 years old, whose Tourette's disorder improved significantly after a period of repeated, sustained practice of activities requiring reflexive responses. One boy engaged in physical exercise including hand-eye co-ordination (table tennis for 6 hours every weekday) and the other learned foreign languages (5 languages within 3 years). Tics may be thought of as a kind of overflow of energy, and excessive energy consumption with physical or mental exercise may improve the motor disorder and associated comorbidities. However, the exercise may require a quick, reflexive response to visual or verbal stimuli. (*Chang Gung Med J 2011;34:650-53*)

Key words: exercise therapy, tic, tic disorder, Tourette's disorder

Tics, the hallmark of Tourette's disorder (TD), are thought of by some sufferers or observers as a state of hyperactivity or an excessive expression of motor energy. Given their semi-voluntary, suppressible nature with premonitory sensations and with complex tics resembling fragments of normal motor programs, they could also be regarded as having a reflex quality.⁽¹⁻⁴⁾ Tics are inhibited by other voluntary motor activity, such as with the use of habit reversal training.^(1,4) However, young children may not be able to engage with or respond well to behavioral therapies. We report 2 children whose tics improved after a period of repetitive and even excessive activities requiring reflexive physical or mental responses.

CASE REPORTS

Case 1

This 11-year-old boy was diagnosed with TD according to DSM-IVTR criteria at the age of 4 years.⁽⁵⁾ The total tic score on the Yale Global Tic

Severity Scale (YGTSS) was 36 at that time.⁽⁶⁾ His mother recalled that his fetal movements were much more active than his siblings. His father had motor tics during elementary school. His mother developed obsessional behaviors related to cleaning at the age of 15 years.

The patient was noted to have frequent blinking, grimacing and throat clearing when he was 3 years old. He was also hyperactive, talkative, and impulsive, although the DSM-IV criteria for attention deficit-hyperactivity disorder (ADHD) were not fulfilled.⁽⁵⁾ His parents never cut his nails because of nail biting. The physical examination was negative for any focal neurological signs. His serum copper concentration was 165 µg/dL (normal, 70-140 µg/dL). The tics seemed to get worse when he ate chocolate. A copper controlled diet was thus instituted and his serum copper concentration returned to a normal level (126 µg/dL) when he was 8 years old.

In elementary school, he received intensive table-tennis training each weekday. Every morning, he played table-tennis from 6 to 9 am before joining

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ordinary class activities. After school, he played table tennis for a further 3 hours. He never complained of being exhausted although his daily schedule was extremely demanding. After 6 months of this regime, his tics decreased dramatically. Furthermore, his nail biting stopped during the following 2 years. His school performance and social activities are both now good. His total tic score on the YGTSS fell to 5 and TD no longer affects him.

Case 2

A 7-year-old boy was diagnosed with TD according to DSM-IVTR criteria at the age of 4 years.⁽⁵⁾ The total tic score on the YGTSS was 50. The patient was noted to be hyperactive prenatally. In infancy, he rarely lay quietly when he was awake, never slept for more than one hour, and cried a lot day and night. When he started to walk independently at his age of 10 months, he liked to help his mother do housework. As he grew up and had his first single word expression at the age of 13 months, his activity increased. His mother tried to severely restrict his physical overactivity during this period but when he was 3 years old, she found it impossible to get him to sit quietly. Six months later, he started to have frequent blinking, grimacing, head nodding and tilting, sniffing and throat clearing, and consistently had tics by the age of 4 years. Increased daytime urinary frequency (pollakiuria) waxed and waned, although a physical examination, urinalysis, and ultrasonographic examination did not show any genitourinary abnormalities.

His parents incidentally found that his tics improved when he was concentrating on learning English, a foreign language for him, especially when he was being tested on English speaking. They started to speak English to him whenever his tics increased. His tics appeared to increase again to an extent within 6 months after he became fluent in English. The parents decided to teach him another foreign language to treat his tics. He cannot now fall asleep unless he listens to language learning tapes, and familiar tapes do not appear to have a good effect. Now at 7 years old, he has quite a good level of spoken English, French, German, Spanish, and Cantonese, as well as his native Mandarin. His tics improved and the total tic score on the YGTSS diminished to less than 30. Pollakiuria disappeared a year ago after two short periods of recurrence. At his

last visit the boy asked the author to prohibit patients with TD from watching TV or playing video games. He felt that these activities were not challenging enough and would aggravate the symptoms of TD.

DISCUSSION

Tics are defined as stereotyped, repetitive, involuntary movements or phonations. However, many individuals with TD also have inattention, hyperactivity, impulsivity, obsessive thinking, and compulsive and self-injurious behavior.⁽⁷⁾ Pollakiuria has been rarely reported although is not uncommon in our experience.^(8,9) The fundamental pathophysiology of TD is still not known, but there is now general agreement that it probably involves dysfunction of the frontal cortico-striato-thalamo-cortical (CSTC) circuitry.^(1,10) The three major groups of symptoms associated with TD as shown in our two cases (tics, obsessive-compulsive disorder, and ADHD) all involve impaired inhibition of unwanted behavior as a common feature.^(10,11) Clinical anecdotes suggest that learning or habit may be involved in the development or maintenance of specific tic behaviors.⁽¹²⁾ Perhaps motor routines are overdone in TD subjects, because of either heightened stress reactivity and awareness of muscular tension, or an intrinsic abnormality in the neural basis of learning these stereotyped fragments of motor behavior.

It has been noted that hyperactivity may develop before the obvious onset of tics.^(1-4,7) In the two cases reported here, signs of hyperactivity appeared as early as the fetal period, although one cannot discount maternal recall bias. It is interesting that in case two, the tics appeared after his daily physical activity was severely restrained by parental dictate, until this became impossible. It may be explained by the natural history of TD or merely coincidental, however possible changes in basal ganglia output and the CSTC circuits induced by physical restraint may potentially play a role.

Serum copper or ceruloplasmin is investigated in children with suspected TD, although the authors are aware of no case of a diagnosis of Wilson's disease in a person appearing to have the possibility. Hypocupremia was reported in one study of adult TD.⁽¹³⁾ We do not know the relation between high serum copper and tics. Some patients claim to have sudden exacerbations of tics after ingestion of a tiny

piece of chocolate, which has a high copper content.

The natural course of TD exhibits improvement in adolescence with severe adult cases being the exception.⁽¹⁻⁴⁾ This makes intervention particularly difficult to assess in small numbers of patients. However, we feel in principle that repetitive practice of physical or mental activities unrelated to the manifestations of TD may be helpful for tics, without the need for the currently recommended management.⁽¹⁴⁾ The symptoms in our two patients were apparently satisfactorily controlled by long-standing learning of a physically skillful, rapid exercise or the acquisition of foreign languages. Playing table-tennis and learning foreign languages seem to be quite different activities. However, both require the learning of a quick, reflexive response to visual or verbal stimuli. The potential effects of similar reflexive exercises warrant further investigation alongside the current resurgence in interest in behavioral treatments for TD.

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需反射反應的重複練習活動於妥瑞症的可能治療角色

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11 歲和 7 歲的兩位男生，他們的妥瑞症在一段時間的需反射反應的重複練習活動後，獲得明顯改善：分別是手眼協調的體能運動（每周五日各六小時的桌球）和學說外國語言（三年中學會說五種語言）。抽動可視為一種能量過多的表現，以對視覺或語言刺激需快速反射反應的體能運動或心智活動大量消耗能量，或許有助於改善妥瑞症的抽動和共病症。（長庚醫誌 2011;34:650-53）

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