Health-related Physical Fitness Management for A Child with Tourette Syndrome

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Tourette syndrome (TS) is a neurobiological disorder characterized by tics, which are repetitive, stereotypical, involuntary movements and vocalizations. Although the causes of TS are not completely understood, previous studies indicated that many children with TS not only experience comorbid conditions such as a lack of concentration, hyperactivity, impulsive behavior, and obsessive-compulsive symptoms, but also demonstrate poorer motor skills than their peers with typical development. There is limited information on health-related fitness exercises and/or physical therapy for children with TS. A 12-year-old boy with TS demonstrated below-age appropriate motor function with a less optimal musculoskeletal condition, including pain in the posterior portion of both lower extremities. After performing individualized health-related physical fitness exercises, he demonstrated a better musculoskeletal condition and motor function. Unexpectedly, he reported he was able to suppress motor tics occasionally by doing stretching exercises. The results suggest that evaluation and management of health-related fitness may be helpful for children with TS. These promising results warrant further investigation of the impact of health-related physical exercises on children with TS. *(Chang Gung Med J 2011;34(6 Suppl):4-9)*

Key words: Tourette syndrome, physical therapy

Tourette syndrome (TS) is a childhood-onset neurobiological disorder characterized by tics, which are repetitive, stereotypical, involuntary movements and vocalizations.[1,2] TS is more common than was previously estimated; approximately 1% of children aged 5–17 years have TS, and 3–6% have tics but do not have all criteria for a diagnosis of TS.[3] Children with TS frequently experience the negative influences of tics and comorbid conditions,[4,5] and also demonstrate health-related fitness problems with fine motor skills,[6-8] insufficient balance control,[9-11] and physical pain due to tics or other unknown reasons.[12] Despite evidence showing that children with TS demonstrate poor health-related quality of life[13] and physical functioning,[6-11] interventional strategies for these children frequently focus only on decreasing tic severity and/or improving behaviour and emotional control, and pay little attention to the children's health-related physical fitness. There is, to our knowledge, no report that describes the effectiveness of health-related fitness exercises and/or physical therapy in children with TS. Therefore, the aim of this case report is to describe an integrated physical therapy program with an emphasis on health-related physical fitness for a child with TS.

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CASE REPORT

A boy with TS was recommended for physical therapy for the first time when he was 12 years old by his pediatric neurologist to improve his health-related physical fitness. He was diagnosed with TS when he was about 7 years old. No other related comorbid diseases were noted. He took vitamin B6 regularly and received anti-tic medication occasionally. The severity of his tics was moderate according to the Yale Global Tic Severity Scale.[1] His body mass index was 29.3 (classified as overweight). The chief complaint of the child was pain in the posterior portion of his lower extremities. He felt an ache and tightness, especially after sudden squatting and jumping, such as when playing basketball. His mother's primary concern was the negative influence of tics in his everyday performance.

Because of the pain in the lower extremities, physical therapy examinations also included a visual analogue scale (VAS) for pain, passive range of motion (ROM), and manual muscle testing of the lower extremities,[14] in addition to the regular physical fitness test which included body composition, cardiovascular fitness, flexibility, muscular endurance, and strength.[15] The movement assessment battery for children (M-ABC), which assesses gross and fine motor function, was used to identify whether this child demonstrated age-appropriate motor functions. In order to understanding his overall health-related quality of life during daily activities, the proxy-responded Child Health Questionnaire, Parent-Form 50 (CHQ PF-50) was completed by his mother. After the above examinations, we identified that the child had potential balance problems; hence, the one-leg standing test was added. Plantar pressure measurements during the one-leg standing test were monitored using a computerized force platform (RSscan International, Olen, Belgium) (Figure).

Evaluations revealed the following impairments in his body function and structure. (1) The child was overweight. (2) The severity of his tics was moderate. (3) The pain in his lower extremities after exercise was rated 6.5 on the VAS, which appeared to be associated with limited ROM of bilateral knee extension and ankle dorsiflexion with a hard end-feel (Table). (4) He showed poor static balance control (one-leg standing test: right leg with eyes open, 10s; left leg with eyes open, 8s; right leg with eyes closed, 5s; left leg with eyes closed, 3s). With respect to the domains of activities and participation, the following observations were made. (1) The child showed below age-appropriate motor ability. (2) Parental stress was endorsed. The mother perceived the child's tics as problematic because they interfered with his functioning. Many arguments between mother and child were observed during this episode of care, especially in the first month. Parenting stress and the child's poor psychological health were con-

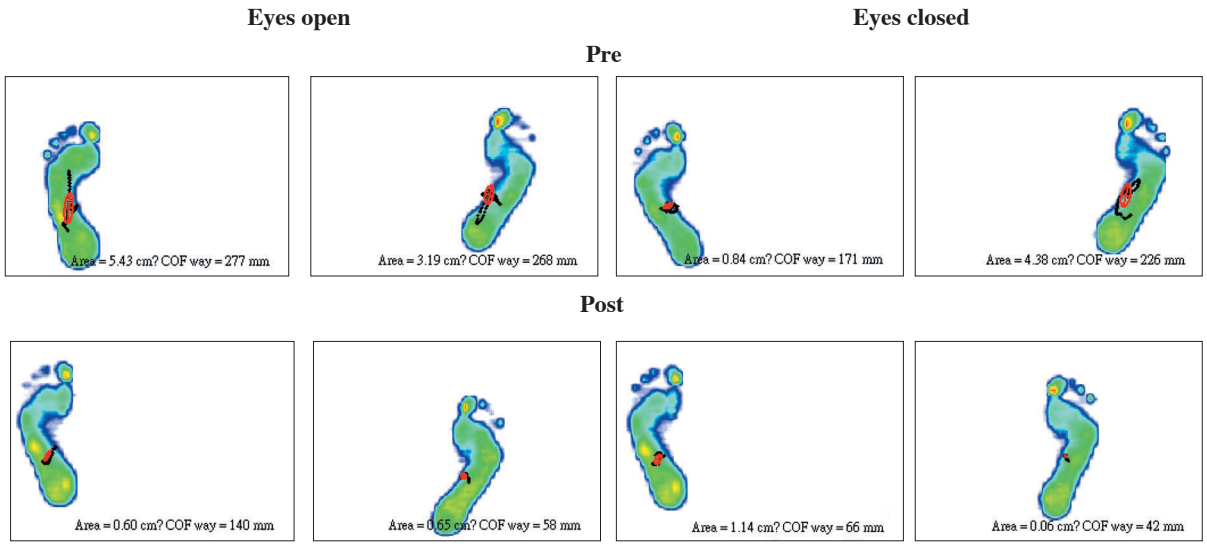

Figure Pre- and post-training comparison of center-of-pressure excursion on a one-leg standing test.

Table Pre- and Post-Training Comparisons of Physical Therapy Examination and Other Tests

	Pre-test	Post-test
YGTSS	37	28
VAS (Pain scale) after exercise	6.5	0
Passive ROM (degrees)		
● Popliteal angle R	0~52°	0~57°
● Popliteal angle L	0~54°	0~57°
● Ankle dorsiflexion R	0~17°	0~20°
● Ankle dorsiflexion L	0~20°	0~25°
M-ABC (Scale score)		
● Manual dexterity	12	8
● Ball skills	3	2
● Static and dynamic balance	10	7
Total impairment score	25	17
Percentile	< 1%	2%
CHQ-PF 50		
● Global health	60.0	85.0
● Physical functioning	83.3	100.0
● Role/social-emotional/behavioral	66.7	100.0
● Role/social-physical	66.7	100.0
● Bodily pain	40.0	80.0
● Mental health	50.0	70.0
● Self- esteem	66.7	75.0
● General health perceptions	43.3	55.8
● Parental impact-emotional	0.0	25.0
● Family activities	58.3	62.5
● Family cohesion scores	60.0	85.0
Physical summary	34.5	52.4
Psychosocial summary	31.5	34.5

Abbreviations: YGTSS: Yale Global Tic Severity Scale; VAS: visual analogue scale; ROM: range of motion; R: right; L: left; M-ABC: movement assessment battery for children; CHQ-PF 50: Child Health Questionnaire, Parent-Form 50.

firmed with the scores on the parental impact-emotional and psychological summary of the CHQ-PF 50 (Table).

Physical therapy intervention was a 2-hour training session conducted once a week for 3 months. This program included (1) aerobic exercise training with treadmill running for 20 minutes, (2) stretch exercises to lengthen the hamstrings and calf muscles, (3) general muscle strengthening with sling exercise therapy with emphasis on closed kinetic chain training, (4) balance training including visual center-of-pressure (COP) feedback training using the same computerized force platform (RSscan International) and walking training on a balance beam, and (5) coordination exercises for the upper extremities. In addition, the child was instructed to perform muscle strengthening, stretching and fine motor skill exercises daily at home.

After the 3-month training, the child reported that pain in the posterior portion of his lower extremities was reduced. His flexibility was improved with increased ROM on knee extension and ankle dorsiflexion (increased angle and normal firm end-feel) (Table). He showed improved balance (one-leg standing test: right leg with eyes open, 24s; left leg with eyes open, 16s; right leg with eyes closed, 22s; left leg with eyes closed, 12s). Pre- and post-training comparisons of COP excursion in the one leg standing test suggested a significantly decreased postural sway (Figure). Overall, his motor abilities and health-related quality of life improved as demonstrated by decreased impairment scores on the M-ABC and increased scores on the CHQ-PF50, respectively (Table). Furthermore, the child demonstrated better writing skills and considered himself a better basketball player. Unexpectedly, the severity of his tics decreased. The child reported that he frequently performed stretching exercises to inhibit tics.

DISCUSSION

In this case report, a thorough physical therapy evaluation was performed in a child with TS. After individualized physical therapy intervention, including balance training and a physical fitness exercise program involving regular aerobic exercise and strengthening, as well as specific stretching, the child's health-related physical fitness and motor functions improved. After the 3-month training, the child reported a reduction in the frequency of tics, reduced pain in the posterior portion of his lower extremities, and the ability to sense the onset of the tic. He also mentioned that he performed stretching exercises to inhibit tics. The results suggest that physical therapy evaluation and management of health-related fitness may be helpful for children with TS.

It is not surprising that a child who has had physical therapy has improved physical-fitness. But unexpectedly, our patient had a reduction in the

severity of tics without taking anti-tic medication. It is well known that in children with TS, tics wax and wane in severity. The presence of tics is also sensitive to a number of factors, including routine psychosocial stress, anxiety, emotional excitement, and fatigue.[4,16,17] The reduction in the severity of tics in this child might have occurred because of better interaction between the mother and child, as shown by the improved scores in the parental impact-emotional domain and the child's psychosocial summary on the CHQ-PF50. Long-term follow-up is required to determine the long-term effects of this 3-month training period on tic severity in this child. According to Banaschewski et al., most children with TS are able to sense when a tic is going to occur, which is called a premonitory urge.[18] This child found his own solution to deal with premonitory urges. It would be valuable to examine whether stretching exercises can help suppress tics in other children with TS.

Although pain is not generally recognized as a symptom of tic disorders, Riley and Lang identified several types of pain as symptoms. These included pain from the actual occurrence of a tic, pain from voluntary efforts to suppress a tic, and pain that is deliberately incited by the individual to obtain relief from a tic.[12] The pain in this child was probably due to the tightness of this muscles. However, the reduction of tics and pain occurred simultaneously, even though the site of tics was not similar to the site of the pain. It is unknown whether the child's tic severity was reduced because of reduction in pain in the lower extremities after physical therapy. It would be interesting to examine the relationship between pain and tics in more detail.

This child's less optimal health-related physical fitness caused several problems, including, poor balance control, coordination and musculoskeletal condition. Because of the complexity of this child's condition, we not only prescribed common exercises, but also added individualized therapeutic excises to deal with his specific impairments, such as poor balance and coordination. The results of this case report cannot determine the effectiveness of common health-related physical fitness exercise in children with TS. There is a need to investigate TS-specific health-related physical fitness problems, as well as the effectiveness of TS-specific health-related physical exercise.

As in other pediatric populations, improved motor skills were demonstrated after physical therapy in this child. However, the motor development of children is not systematically examined in the school environment in Taiwan. Therefore, this child's delayed motor development was not identified until this episode of care. It is not known whether late identification of motor delays contributes to tic severity or whether tics interfere with the development of advanced motor function in a child. These questions need to be examined further to understand how to promote optimal health-related physical development and fitness in children with TS. In conclusion, the results of this case report suggest that evaluation and management of health-related physical fitness may be helpful for children with TS. These promising results warrant further investigation of the impact of health-related physical fitness exercises in children with TS.

REFERENCES

1. Leckman JF. Tourette's syndrome. Lancet 2002;360:1577-86.
2. Storch EA, Lack CW, Simons LE, Goodman WK, Murphy TK, Geffken GR. A measure of functional impairment in youth with Tourette's syndrome. J Pediatr Psychol 2007;32:950-9.
3. Eapen V, Crncec R. Tourette syndrome in children and adolescents: special considerations. J Psychosom Res 2009;67:525-32.
4. Swain JE, Scahill L, Lombroso PJ, King RA, Leckman JF. Tourette syndrome and tic disorders: a decade of progress. J Am Acad Child Adolesc Psychiatry 2007;46: 947-68.
5. Bloch MH, Peterson BS, Scahill L, Otka J, Katsovich L, Zhang H, Leckman JF. Adulthood outcome of tic and obsessive-compulsive symptom severity in children with Tourette syndrome. Arch Pediatr Adolesc Med 2007;160: 65-6.
6. Bloch MH, Sukhodolsky DG, Leckman JF, Schultz RT. Fine-motor skill deficits in childhood predict adulthood tic severity and global psychosocial functioning in Tourette's syndrome. J Child Psychol Psychiatry 2006;47:551-9.
7. Prodoehl J, Corcos D, Vaillancourt D. Basal ganglia mechanisms underlying precision grip force control. Neurosci Biobehav Rev 2009;33:900-8.
8. Nowak D, Rothwell J, Topka H, Robertson M, Orth M. Grip force behavior in Gilles de la Tourette syndrome. Mov Disord 2005;20:217-23.
9. Albin RL, Mink JW. Recent advances in Tourette syn-

drome research. Trends Neurosci 2006;29:175-82.
10. Lemay M, Termoz N, Leperance P, Chouinard S, Rouleau GA, Richer F. Postural control anomalies in children with Tourette syndrome. Exp Brain Res 2007;179:525-30.
11. Stinear C, Coxon J, Byblow W. Primary motor cortex and movement prevention: Where Stop meets Go. Neurosci Biobehav Rev 2009;33:662-73.
12. Riley DE, Lang AE. Pain in Gilles de la Tourette syndrome and related tic disorders. Can J Neurol Sci 1989;16:439-41.
13. Elstner K, Selai CE, Trimble MR, Robertson MM. Quality of life (QOL) of patients with Gilles de la Tourette's syndrome. Acta Psychiatr Scand 2001;103:52-9.
14. Chou PC, Chen YY, Lin YH, Lien HY, Hwang WB, Liu WY. Lower extremity musculoskeletal measures in children: reliabilities and relationships to balance functions. Formosan J of Physical Therapy 2007;32:217-25.
15. Corbin CB, Pangrazi RP, Franks BD. Definitions: Health, fitness and physical activity. Pres Counc Phys Fit Sports Res Dig 2000;3:1-8.
16. Findley DB, Leckman JF, Katsovich L, Lin H, Zhang H, Grantz H, Otka J, Lombroso PJ, King RA. Development of the Yale children's global stress index (YCGSI) and its application in children and adolescents with Tourette's syndrome and obsessive compulsive disorder. J Am Acad Child Adolesc Psychiatry 2003;42:450-7.
17. Hoekstra PJ, Anderson GM, Limburg PC, Korf J, Kallenberg CG, Minderaa RB. Neurobiology and neuroimmunology of Tourette's syndrome: an update. Cell Mol Life Sci 2004;61:886-98.
18. Banaschewski T, Woerner W, Rothenberger A. Premonitory sensory phenomena and suppressibility of tics in Tourette syndrome: developmental aspects in children and adolescents. Dev Med Child Neurol 2003;45:700-3.

對一名妥瑞症兒童的健康相關體適能處理

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妥瑞症 (Tourette syndrome) 是一種神經生物學上的疾病，主要會出現重複的、刻板性的、半不自主的動作及聲音上的抽動 (tic)。在病因方面，目前雖尚無定論，但過去的研究指出，許多妥瑞症兒童不僅合併了許多共病症，例如注意力不足、過動與衝動行爲以及強迫症狀；在動作技巧上也較同年齡的正常發展兒童表現來的差。然而，針對妥瑞症兒童的健康相關體適能 (health-related physical fitness) 及／或物理治療的相關資訊仍相當有限。因此，本個案報告的目的即爲描述對一位妥瑞症兒童所實行的一個整合性的健康相關體適能處理程序。一名 12 歲的妥瑞症男孩有低於年齡應有的動作功能和較不良骨骼肌肉系統，如雙下肢後側疼痛。在實施了個別化健康相關體適能運動訓練後，這位兒童的骨骼肌肉系統問題和動作功能獲得到改善。出乎意料地，這名兒童感覺他可藉由施行伸展運動 (stretching exercises) 抑制 tic。本報告結果建議健康相關體適能的評估與處理也許對妥瑞症兒童有所幫助。這些效果顯示未來值得針對健康相關體適能運動對於妥瑞症兒童的影響進行研究。(長庚醫誌 2011;34(6 Suppl):4-9)

關鍵詞：妥瑞症，物理治療

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