Motor ability and visual-motor integration in children affected by tic disorder

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Summary

Background: previous studies have shown that patients with Tourette Syndrome (TS) present deficits in visual-motor integration, fine motor skill and spatial/motor ability. More recently, it has been suggested that fine motor skill deficits may be predictors of future tic severity and global psychosocial functioning in TS children.

Objective: we hypothesized that deficits of motor coordination could be present in a large number of children with Tic Disorders (TD) and this could constitute a further source of social impairment. With this purpose, we performed a cross sectional evaluation of motor coordination and visual-motor integration in a large group of TD children.

Methods: our sample was constituted by 96 TD children (19 females, 77 males) with an age range between 4 and 13 years. 18 children presented a diagnosis of Transient Tics, 16 of Chronic Multiple Motor or Phonic Tics and 62 of Tourette Syndrome. Exclusion criteria were the presence of a diagnosis of mental retardation or autistic spectrum disorder; all patients were drug naive. Patients underwent a focused neuropsychological testing battery of Movement Assessment Battery for Children (M-ABC) and Beery-Buktenica Developmental Test of Visual-Motor Integration (VMI). We used for the statistical analysis ANOVA univariate test, t-test and chi-square test.

Results: we have observed, in a large part of our sample, difficulties in motor ability. 19% of children obtained pathological scores (inferior to the 5th percentile) on the total M-ABC and another 17% achieved borderline scores (between the 5th and 15th percentile); the higher rate of impairment was found in the manual dexterity subtests (33% had pathological scores and 22% borderline scores). No association was found between the categorical M-ABC total scores (categorized as normal/borderline/pathologic, or grouping together borderline and pathologic) and TD diagnosis or tic severity measured by YGTSS. Children with higher YGTSS scores tended to show poorer performances in M-ABC, in particular on the manual dexterity subitems, but the correlation did not reach a statistical significance.

Conclusions: our results confirmed the clinical impression that many children with TD are clumsy. In further studies, two aspects deserve to be expanded, the developmental trajectories of children showing poor motor performance and the relationship of motor disabilities with other problems (ADHD, learning disabilities) that impair a large set of children affected by TD.

KEY WORDS: tic disorder, Tourette syndrome, motorability, visual-motor integration, DCD.

Background

Tic disorders are common in childhood and adolescence and their prevalence is about 22.3% in preschoolers (3-5 years) and 3.4% in adolescents (12-18 years) (1). The classifications of the disorders range from Transient Tic Disorder to Tourette Syndrome (TS), according to the type of tics and to the duration of the symptoms. TS is a neuro developmental disorder characterized by childhood onset, multiple motor tics and one or more vocal tics over the course of at least 1 year. Its overall prevalence is about 1% in youngsters aged 5-18 (2). Basal ganglia dysfunction and the related corticostriato-thalamo-cortical circuits are thought to be involved in the pathogenesis of the disorder. TS is commonly associated with comorbid disorders, such as attention-deficit hyperactivity disorder (ADHD), obsessive-compulsive disorder (OCD) or obsessive and compulsive behaviours (OCB), depression, anxiety disorders, learning disabilities (LD), explosive outbursts of aggressive behaviour. Several Authors have focused on executive functions in children with TS (3) but definitive data are uncertain, especially because of the high overlap of the impairments between TS and different comorbidities, such as ADHD. Previous studies conducted highlight that children with TS can show difficul-
ties in the areas of fine motor skills, handwriting, and visual-motor integration. Incagnoli and Kane (4) found consistent and specific problems in tasks requiring visual-graphic abilities in 13 boys with TS between 10 and 13 years of age; these data suggested the presence of a dysfunction of non-constructual visual-practical abilities. Bornstein et al. (5) evaluated a sample of 28 subjects with TS, aged 7-23, all medication-free. They used psychological and neuropsychological tools. Motor and visual-motor abilities were tested with the Finger Tapping Test, the Grooved Pegboard Test and the Reitan-Klove Sensory-Perceptual Examination. Compared with available normative data, the frequency of abnormal test performances was variable, but more frequent in motor and sensory tasks. Brookshire et al. (6) compared neuropsychological characteristics of TS children with a control group of normal siblings and with another group of patients with arithmetic disabilities (AD). TS group showed poor performance on written arithmetic tasks, like also the AD group; but only TS group showed difficulty in visual-motor task that included copying or drawing with a pencil. Schuerholz et al. (7) analysed neuropsychological data from 3 groups of children formed by TS-only, TS + ADHD, TS with clear ADHD characteristics but not ADHD diagnosis. In their analyses the Authors highlighted that the TS-only group performed better the tasks requiring visual-spatial skill. In another study (8), neuro-motor functions were assessed using the timed tasks from the PANESS, in 3 groups of children formed by TS only, TS + ADHD, AD-HD only. The TS only group performed the fastest movements, bolstering the authors' hypothesis that children with TS do not exhibit motor difficulties and slower execution of the movements. But interestingly, the slowest movements were displayed by the group of TS + ADHD. Schultz et al. (9) proposed and tested a neuropsychological model of visual-motor integration skill in children with TS and healthy control children. Children with TS performed significantly worse than control children in the Beery-Buktenica Visual-Motor Integration (VMI) Test. Consistent with the proposed model, visual-perceptual abilities, measured with Bloch Design of the WISC-R and Matrice subtest of the Kaufman Brief Intelligence Test, and fine-motor coordination sub-processes, measured with Purdue Pegboard, were significant predictors of VMI scores. No significant correlation was found between VMI performances and sustained attention or motor inhibition. Both the above-mentioned studies seem to demonstrate the underlying involvement of the caudate nucleus, basal ganglia and fronto-cortico-striatal circuits in the pathogenesis of TS, rather than investigating the motor abilities of TS children. More recently, Bloch et al. (10) have suggested that fine motor skill deficits may be predictors of future tic severity and global psychosocial functioning in children with TS. We have hypothesized that deficits in motor ability could be present in a large number of children affected by Tic Disorders and that this might constitute a further source of social impairment. For this purpose, we performed a cross sectional evaluation of motor ability and visual-motor integration in a large series of children affected by Tic Disorders.

Methods

Participants

96 TD children (19 females; 77 males) were recruited from the Unit of Movement Disorders of the Child and Adolescent Neuropsychiatry Department, “Sapienza” University of Rome, Italy. 18 presented Transient Tics, 16 a diagnosis of Chronic Multiple Motor or Phonic Tics and 62 a diagnosis of Tourette Syndrome; their age ranged between 4.1 and 12.9 years, median: 9.1. All the subjects had a comorbidity diagnosis of mental retardation or autistic spectrum disorder were excluded from our sample. The pathogenesis of tics was not due to neurodegenerative diseases or structural brain lesions of vascular, traumatic or inflammatory origin. All children attended normal schools and only 4 of them had special education programs. All subjects were at their first neuropsychiatric evaluation and all were drug naïve. All patients were initially assessed by semi-structured diagnostic interviews and self-report inventories. Their tic severity, measured by the YGTSS, ranged between 4 and 52 (mean: 20.59). The patients underwent a focused neuropsychological testing battery consisting of Movement Assessment Battery for Children (M-ABC) and Beery-Buktenica Developmental Test of Visual-Motor Integration (VMI). Their parents compiled the Children Behaviour Checklist (CBCL). To minimize the possible interference of tics on the motor performance, all neuropsychological tests were held during the morning. Subjects were recruited only after the informed consensus of their parents. The study was approved from the Institutional Review Board of the Department.

Instruments

The Movement Assessment Battery for Children (M-ABC) (11) is specifically developed for the identification and evaluation of children with mild to moderate motor impairment. The M-ABC is organized into three domains or performance areas, covering manual dexterity (3 items), ball skills (2 items), static (1 item) and dynamic balance (2 items). A special feature of the M-ABC test is the organisation in four different age bands between 4 and 12 years. Visual perceptual skills were assessed using the VMI and the VMI Supplemental Developmental Tests, Visual Perception and Motor Coordination (12). The Beery VMI is a copy forms test that is frequently administered during visual perceptual evaluations. It is a well-researched, commonly used, standardized test with an objective scoring system. The VMI supplemental tests contain the same forms, specifically designed to identify visual analysis and motor coordination difficulties. In particular, the Supplemental Developmental Test of Visual Perception assesses the child’s visual analysis/visual spatial skills in a motor-reduced fashion. On the other hand, the Supplemental Developmental Test of Motor Coordination test focuses on the motor integration skill of the patients and minimizes the analysis requirement. The Child Behaviour Checklist (CBCL) (13) is designed to obtain parental reports of the child.
emotional and behavioural problems in a standardized format, which provides normative data. Scores equal to or above the 98th percentile for controls (T score above 70) are considered abnormal. We analysed M-ABC, its sub-items and their corresponding percentile scores as discrete variables and as ordinal variables using the test developer’s normative cut-offs. VMI and its supplementary tests were analysed as discrete variables (using the standard scores) and as ordinal variables using the test developer’s normative cut-offs. We used for the statistical analysis ANOVA univariate test, t-test and chi-square test, as appropriate. The value of p<0.05 was considered statistically significant.

Results

Our results showed motor impairments in several patients. Particularly, 19% of children obtained pathologic scores (inferior to the 5th percentile) on the total M-ABC and another 17% achieved borderline scores (between the 5th and 15th percentile); the higher rate of impairment was found in the manual dexterity subtests (33% had pathological scores and 22% borderline scores), while static and dynamic balance subtests (respectively 9% and 20%) and ball skills subtests (11% and 10%) were less affected. Total VMI was abnormal in 16% of children, Motor Coordination subtest in 17% and Visual Perception subtest in 5% of all patients. There was a significant association between the results of the two neuropsychological tests. In fact ANOVA univariate analysis showed a significant result between the categorized (normal, borderline and pathologic) total scores of M-ABC and the results of Motor Coordination (F=5.312; p=0.007) and of Visual Perception (F=5.228; p=0.007) VMI supplementary tests. These results were confirmed by chi-square test between M-ABC categories (normal, borderline, pathologic) and normal/pathologic score of the VMI supplementary tests, Motor Coordination (χ2 = 11.121; p= 0.004) and Visual Perception (χ2 = 8.401; p=0.015). We analysed our data grouping together the patients who obtained pathologic and borderline total M-ABC scores and comparing them to children who obtained normal M-ABC scores. T-test confirmed the significant correlation between the categorized scores of M-ABC and the results of Motor Coordination and of Visual Perception VMI supplementary tests (Tab. 1). The results were confirmed by chi-square test between M-ABC categories (normal versus borderline plus pathologic) and normal/pathologic scores of the VMI supplementary tests, Motor Coordination (χ2 = 5.8; p=0.016) and Visual Perception (χ2= 8.386; p=0.004). No association was found between the categorical M-ABC total scores (categorized as normal/borderline/pathologic) and Tic Disorder’s diagnosis or tic severity as measured by YGTSS. Children with higher YGTSS scores showed poorer performances in M-ABC, in particular to the manual dexterity sub-items, but the correlation did not reach a statistical significance. The CBCL parent’s reports disclosed that Total T was abnormal in 28% of subjects, Internalization T in 28% and Externalization T in 13%. A series of CBCL subscales were abnormal in several children: Withdrawn in 13%, Somatic Complaint in 19%, Anxious/Depressed in 28%, Social Problems in 16%, Thought Problems in 14% and Attention Problems in 18%. Moreover, ANOVA univariate analysis showed a significant association between M-ABC scores (categorized as normal/borderline/pathologic) and some CBCL subscales: Withdrawn, Internalization, Total T, Though Problems, Anxious/Depressed and Attention Problems (Tab. 2). These associations continue to be significant also grouping together patients who obtained borderline and pathologic total M-ABC scores (Tab. 1).

Discussion

According to our hypothesis, in this sample we have observed that a large proportion of our children showed deficits in motor ability. In fact near 50% of TD patients obtained pathologic (<5th percentile) or bor-

| Table 1 - T-test. Significant associations between the results of categorized M-ABC total scores (grouping together borderline and pathologic versus normal values) and the result of VMI and CBCL. |
|---|---|---|
| VMI | Motor Coordination | 3.009 |
| | Visual Perception | 2.300 |
| | Total T | -2.588 |
| | Internalization | -3.066 |
| | Withdrawn | -2.655 |
| | Anxious/Depressed | -2.484 |
| | Social problems | -2.473 |
| | Thought problems | -2.393 |
| | Attention problems | -2.109 |

| Table 2. ANOVA univariate analysis. Significant associations between the results of M-ABC total scores (categorized as normal, borderline and pathologic) and the result of VMI and CBCL. |
|---|---|---|
| VMI | Motor Coordination | 5.312 |
| | Visual Perception | 5.228 |
| | Total T | 3.346 |
| | Internalization | 4.179 |
| | Withdrawn | 4.535 |
| | Anxious/Depressed | 3.314 |
| | Thought problems | 3.412 |
| | Attention problems | 3.093 |
derline (between 5th and 15th centile) total scores in the MABC. It should be stressed that a cut-off at the 15th centile is proposed in clinical practice for assessment of motor problems (14). The difficulties in motor performance presented in our patients are similar to those found in children affected by the disorder known over the years under different names, such as “clumsy child syndrome”, “minimal brain dysfunction” or “developmental apraxia” and more recently defined as “Developmental Coordination Disorder” (DCD) (15, 16). The DSM-IV definition of DCD includes that “the disturbance is not because of a general medical condition (e.g. cerebral palsy, hemiplegia, or muscular dystrophy) and does not meet criteria for a pervasive developmental disorder” (17). Some Authors (15) include TD and TS among the medical conditions that should be ruled out before posing a diagnostic of DCD, while others do not (15, 16). It is not the purpose of this work to establish if DCD could be another comorbid condition in TD or TS, even if it is difficult to consider transient or mild tics as a medical condition excludary of a diagnosis of DCD. Our patients show difficulties in different areas but, according to previous studies (3), manual dexterity seems to be the most impaired skill. In particular, in the over-mentioned study of Bloch et al. (10), the poor performance at Purdue Pegboard seems to correlate with the severity of tics at the time of childhood assessment and to predict the adulthood severity of tics. In our sample we did not find any correlation between both global motor and fine motor impairment with the kind of TD; moreover, the children with higher severity tics show a statistically not significant trend to have more manual difficulties. However, due to cross-sectional design of our study, we are not able to confirm the possible predictive meaning of fine motor deficits. We have observed in our patients that the VMI impairment is less frequent than motor impairment; but the association we have found between the performances on the MABC and those in both Motor Coordination and Visual Perception VMI subtests, supports the hypothesis of an impairment of visual-motor integration processes in these children, compared with those that show normal VMI results. In the end, in our sample we have pointed out an association between poor motor performances and several subscales of CBCL parent’s reports, especially internalization and withdrawn behaviours. Children affected by TD or TS may suffer from a large series of psychopathologic comorbidities (among others depression or anxiety), whose relationship with the basic disorder is far from being clarified. In a previous study (18) we found a relationship between the long duration of TD and internalizing problems, probably due to difficulties in social life or to a reduction of self-confidence. On the other hand, children and adolescents with poor motor ability have been identified as being at risk of social, emotional and behavioural problems due to their motor difficulties (19-22). Further studies are necessary to better understand the possible interaction between motor dysfunctions and psychopathological problems that could play an important role in the development of psychosocial impairment in children with TD.

Conclusions

TD and TS are puzzling entities, affecting people in different areas (neurological, emotional, behavioural) with different degrees of impairment. Our results confirmed the clinical impression that many children with TD are clumsy. But on the other hand, individuals affected by TS are reported to have a particular talent in sports or in other activities in which the motor ability is pivotal, as playing music (for example Ray, the protagonist of the famous Sack’s tale) (23). The meaning of this finding should be confirmed by other studies; among others, two aspects deserve to be expanded: the developmental trajectories of children showing poor motor performance and the relationship of motor disabilities with other problems (ADHD, learning disabilities) that impair a large set of children affected by TD. The main limitation of this study is that we have analysed the results of neuropsychological testing of our patients in comparison with normative data. The reason of this choice is that very few reports in this field are present in Literature, and thus this study was conceived as an exploratory investigation to confirm the clinical impression of an impairment of motor ability in children affected by TD and to quantify the extent of this phenomenon. Further studies utilizing control samples are needed to support our data and to deepen our understanding of the mechanisms underlying the motor dysfunctions we have found. On the other hand, its strengths are to have examined a large number of children affected by different kinds of TD at their first neuropsychiatric evaluation and drug naïve, thus giving us the opportunity to perform a quasi-naturalistic observation without any confounding pharmacological intervention.

References

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