

Toe walking in children with autism spectrum disorder and comorbid attention deficit hyperactivity disorder: An early marker of a heterogeneous disorder?

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ABSTRACT

Autism spectrum disorder (ASD) is a complex neurodevelopmental disorder (NDD). Toe walking has been reported in children with NDDs. However, the relationship between ASD and toe walking is still unclear. The aim of the paper is to describe toe walking in four children with ASD and comorbid attention deficit hyperactivity disorder (ADHD). Their age ranged between 28 months and 42 months and three of the cases had moderate to severe toe walking on both feet. All the cases scored in the high-risk category (8–20) of the Modified Checklist for Autism in Toddlers and above the cut-off scores (24 for girls and 32 for boys) of the ADHD RS-IV-P. In addition, they all met DSM-V diagnostic criteria for ASD and ADHD. Clinicians diagnosing ASD may be omitting an important element of the disorder in the form of toe walking, which could contribute to delayed or inaccurate diagnoses. Consideration of including motor function deficits such as toe walking in the diagnostic criteria for ASD might improve early detection and intervention.

Key words: Attention deficit hyperactivity disorder, Autism spectrum disorder, Diagnosis, Toe walking

Toe walking (also referred to as the tip-toe gait) is a bilateral gait abnormality in which a normal heel strike is absent and most weight bearing occurs through the forefoot with an absence of full foot contact during the entire standing phase [1]. Orthopedic surgeons and physiotherapists describe toe walking in terms of decreased ankle range of motion and inability to heel strike at the initial contact of gait [2]. While toe-walking gait in some children is due to an underlying pathology such as cerebral palsy, there are no obvious explanations in others, and this type is referred to as idiopathic or habitual toe walking (ITW). The abnormality may not be pathologic in children who are <2 years of age. It is a pattern sometimes observed in healthy developing children of <2 years old who are learning to walk independently. However, a persistent toe walking beyond 2 years of age can be the first sign of an upper motor neuron or neuromuscular disease. It has been hypothesized that ITW is a motor expression of minimal cerebral dysfunction.

Autism spectrum disorder (ASD) is a neurodevelopmental disorder (NDD) that affects speech development and social interactions. Toe walking has been reported in children with NDDs [3,4]. For example, a study among children with attention deficit hyperactivity disorder (ADHD) found that 20.8% of them had ITW. Similarly, a prevalence rate of 20% was found in


individuals with ASD [5]. A study that compared 56 children with toe walking and 40 controls found that those with toe walking scored significantly lower on all developmental markers measured in the study [6]. However, the link between ITW and NDDs is not being established and the possibility of ITW being a marker of NDDs, especially ASD has not been explored. This case series is intended to further provide information about this link.

CASE SERIES

We presented a case series of four children. The case series was reported following the recommendations outlined in the CAsEReport (CARE) guidelines. Written informed consent was obtained from the caregivers of the patients for publication of this case series. All four children in this case series were receiving care at a care center. Details about the center are described elsewhere [7]. Other findings on their toe walking are shown in Table 1. They all met diagnostic criteria for ASD and ADHD according to the Diagnostic and Statistical Manual – Fifth Edition (DSM-V). In addition, they were assessed on the Bayley screening test (BST), ADHD rating scale-IV preschool version (ADHD RS-IV-P), and the Modified Checklist for Autism in Toddlers (MCHAT). Details of family, pregnancy, neonatal, and childhood history were obtained from parents of the four children. The children were classified based on the severity of toe walking [8].

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All four children underwent physical examination, during which passive and active ranges of motion in the hip, knee, and ankle joints were measured with a goniometer in the supine position. They were assessed in a quiet, spacious room equipped with all materials necessary for the assessment. Each child was examined wearing only shorts and the back and extremities were inspected. Ankle range of motion was measured with the knee in the extended position. Gait, balance, deep tendon reflexes, muscle tone, and presence of possible Babinski signs were evaluated using standard procedures.

The result of the assessment on BST, ADHD RS-IV-P, and MCHAT tools is shown in Table 2. The diagnosis was made based on DSM-V criteria and the assessment tools above. All the cases were commenced on an average of 30 h/week of therapy that included speech therapy, occupational therapy, and behavioral therapy. All four cases were also started on daily passive stretching of ankle tendon Achilles and the use of ankle-foot orthosis to correct the toe walking.

Case A

Our first case is a 38-month-old male child who was born to a 32-year-old mother through spontaneous vertex delivery (SVD) and he cried immediately after birth. He presented with a history of poor speech development, repetitive behavior of running in circle, hyperactivity, and self-biting. These symptoms were

noticed at 18 months of life. There were no adverse reports during the neonatal period. He is the 2nd of two children. Other aspects of developmental history are in Table 3. On general examination, he was healthy-looking, he was hyperactive, climbing chairs and was attempting to leave the consulting room. His eye contact was poor and did not respond to questions, including greetings. Toe walking was obvious with both feet. He has had five weeks of therapy with minimal improvement in self-biting and hyperactivity. There has not been any significant improvement in toe walking.

Case B

Case B is a 42-month-old male child, 1st of three children. He was born to a 28-year-old mother through SVD following induction of labor as a result of prolonged labor. He did not cry immediately after birth and needed to be resuscitated for about 10 min before he cried. He presented with poor speech development, poor social interaction, and repetitive behavior of rocking on a spot and recurrent generalized seizure. The symptoms started about 20 months of life. Other aspects of developmental history are in Table 3. General examination revealed a male child, he was continuously moving and fidgeting on the chair. He repeatedly throws the items on the consulting table to the floor and moves between sitting on the table and the chair. His eye contact was poor and he had no speech. There was toe walking in both feet, which

Table 1: Sociodemographic and clinical characteristics of cases

Case	Age (months)	Sex	Family history of toe walking	Maximum passive dorsiflexion of ankle (°)		Maximum passive plantar flexion of ankle (°)		*Severity of toe walking (Based on dorsiflexion)	
				Rt Foot	Lf Foot	Rt Foot	Lf Foot	Rt Foot	Lf Foot
A	38	M	Yes	10	10	35	35	Type 2	Type 2
B	42	M	No	15	5	40	45	Type 2	Type 3
C	40	M	No	5	5	40	40	Type 3	Type 3
D	28	F	No	20	20	25	20	Type 1	Type 1

*Classification of severity of toe walking: Type 1=Mild, Type 2=Moderate, and Type 3=severe [8]

Table 2: Scores on assessment tools

Case	Bayley screening score				Gross	ADHD RS-IV-P	MCHAT
	Cognitive	Receptive	Expressive	Fine			
A	25 ^b	18 ^b	14 ^a	18 ^b	22 ^b	45	12
B	12 ^a	13 ^a	8 ^a	12 ^a	20 ^a	41	14
C	26 ^b	15 ^b	15 ^a	26 ^c	24 ^b	38	10
D	23 ^a	12 ^a	7 ^a	14 ^a	15 ^a	42	12

^aAt risk of delay; ^bEmerging risk; ^cCompetent (lowest risk). ADHD RS-IV-P=Attention Deficit Hyperactivity Disorder Rating Scale-IV preschool version (Total score for boys>32, Total score for girls>24, suggest ADHD). MCHAT=Modified Checklist for Autism in Toddlers (Low risk=0-2, Moderate risk=3-7, High risk=8-20)

Table 3: Developmental history

Case	GA (Weeks)	Birth Wt (kg)	Apgar Score (At 5 min)	Onset of selected developmental milestones (in months)						Comorbidity
				Neck control	Standing without support	Walking	First word	Bladder control	Bowel control	
A	38	3.4	8	3	9	11	YTA	36	YTA	Nil
B	40	3.8	NA	4	11	14	YTA	YTA	YTA	Epilepsy
C	40	3.2	9	3	8	11	YTA	YTA	34	Epilepsy
D	39	3.0	9	5	12	18	YTA	YTA	YTA	Epilepsy

GA=Gestational age (weeks); NA=Not available; YTA=Yet to attain

was worse in the left foot. His intervention has been irregular as a result of financial constraints.

Case C

Case C is a 40-months-old male child, he was an only child. He was born to a 30-year-old mother through SVD and was said to have cried immediately after birth. He presented with poor speech development, preference for solitary play, repetitive hand flapping, clapping, and difficulty following instructions that were noticed at 24 months of life. These symptoms were noticed at about 18 months of life. He was also hyperactive and loves to climb objects. Other aspects of developmental history are in Table 3. General examination showed a hyperactive child; he, however, responded to the parent's instructions when asked to sit, though usually short-lived. He attempted to respond to questions, but the words were incomprehensible. His response to therapy has been slow, with no significant improvement after four weeks of therapy.

Case D

Case D is a 28-month-old female child, 2nd of two children. She was born to a 25-year-old mother through SVD and reported to have cried immediately after birth. She presented with poor speech development, preference for solitary play, poor eye contact, hyperactivity, and difficulty sitting still noticed about 6 months before presentation. The symptoms started at 22 months. There was a history of infantile spasm that was diagnosed at the age of 25 months, and she is on medications. Other aspects of developmental history are in Table 3. General examination showed a hyperactive girl, mostly on the go, climbing tables and chairs. Her eye contact was poor and she had no appreciable speech. She made significant improvements in all the affected areas. The maximum passive dorsiflexion of the ankle in both feet increased from 20° to 25° after 4 weeks on ankle-foot orthosis.

DISCUSSION

The four cases in our report met the diagnostic criteria for both ASD and ADHD, and they all had toe walking gait. Previous studies have reported toe walking in children with ASD and ADHD independently. It has, however, been observed that etiological factors that are found in children with developmental disorders are also related to toe walking. For example, toe walking has been associated with different complications occurring during pregnancy, delivery, and the postpartum period [9]. Most cases of ADHD are predominantly a motor problem, so also studies have proposed the hypothesized that autism is primarily a disorder of movement first [10], and there are reported impairment of basal ganglia and cerebellum in ASD [11]. Motor delays seem to be one of the earliest risk markers for ASD and relate to the subsequent severity of social and communication deficits.

The concept of comorbidity, which is defined as the co-occurrence of two or more disorders in the same person, is gaining attention in ASD in the last decade. It has been reported

that children with ASD had more severe comorbid symptoms than atypically developing toddlers. The prevalence of ADHD in those with ASD has ranged from 14% to 78% [12,13]. Individuals with ASD and comorbid ADHD often have poorer outcomes than children with ASD alone [14]. Most research on comorbidity in ASD has focused on psychiatric disorders and often excludes issues related to their ambulatory function such as seen in toe walking.

Early detection and intervention are one of the most important factors contributing to better outcomes in developmental disorders. The current diagnostic criteria for ASD as documented in the DSM-5 do not include movement disorder as a symptom in diagnosing ASD. However, with recent findings in the literature, clinicians diagnosing ASD may be omitting an important element of the disorder in the form of toe walking, which could contribute to delayed or inaccurate diagnoses. Consideration of including motor function deficits such as toe walking in the diagnostic criteria for ASD might improve early detection and intervention.

CONCLUSION

The distinct trajectories of patients with ASD regarding clinical manifestations have been largely limited to neurobehavioral criteria from the Diagnostic and Statistical Manual of Mental Disorders. These neurobehavioral symptoms of ASD often mimic symptoms of other disorders such as language disorders and this often delays early diagnosis. Motor deficit and hyperactivity symptoms are identified early and can, therefore, lead to (early) screening activities and the development of prevention programs. In addition, ASD has been described as a heterogeneous disorder with complex genetic underpinnings, hence investigating further the link between motor toe walking, ADHD and ASD might help to describe a subpopulation of children with ASD.

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