



Can Rhythmical Auditory Stimulation Alter Gait Pattern in Children with Asperger Syndrome?

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Abstract

The aims of this study were to evaluate the gait abnormalities in children with Asperger syndrome (AS) and to investigate the effectiveness of rhythmical auditory stimulation (RAS) on gait training in children with this condition. Five children with AS (mean age: 8.5 ± 2.0 years) and 6 children with normal development (mean age: 9.5 ± 2.0 years) participated in this study. The participants were asked to walk on a treadmill under each of the following conditions conducted in sequence: (1) walk at a comfortable speed with no RAS (baseline), (2) walk at a comfortable speed accompanied by RAS (sound-on condition), and (3) walk again at a comfortable speed with no RAS (sound-off condition). The temporal and kinematic gait parameters of the walking in each condition were recorded with a VICON 370 system. No significant differences were found among the baseline, sound-on, and sound-off gait parameters in either group. The children with AS rotated their pelvis more during walking than the control group did at baseline ($p=0.018$) and during the sound-on ($p=0.011$) condition. Moreover, the control group spent less time in the double-leg supporting position in the sound-on and subsequent sound-off condition than the AS group did. No statistically significant differences were found between the two groups in all other gait parameters (i.e., step length, step width, step height, hip and knee joint angles at mid-stance phase of gait) in the three testing conditions. The children with AS demonstrated excessive pelvic rotation during walking when compared to children with normal development. A short period of gait training with RAS might not be able to improve the walking pattern in children with AS.

Keywords: Autism spectrum disorder; Sound cue; Walking pattern; Gait training; Movement disorder

Introduction

Asperger syndrome (AS), a form of autism spectrum disorder, is a fairly common condition among children and youth. It is estimated that as many as 48 per 10,000 children have this disorder [1]. According to the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV-TR), children with AS are characterized by impairment in social interaction, occupation, or other functions [2]. They also demonstrate repetitive and restricted stereotyped patterns of behavior, activities, and interests [2]. Although AS is foremost a social disorder, sensorimotor impairments are widely reported in this group of children [3-5]. Wing first reported that children with AS tend to have poor motor skills, coordination, and balance problems that may affect their daily function, sporting skills, and writing ability [6,7]. Using the standardized Bruininks Oseretsky Test of Motor Proficiency [8], Ghaziuddin and Butler also reported that gross motor skills such as running speed and agility are inferior in children with AS when compared to the norm [3].

Walking is amongst the most important and fundamental motor skills that a child needs to acquire during the first few years of life [9]. It has been reported that although children and adults with autism can walk independently [4,10], they demonstrate atypical gait characteristics such as decreased knee flexion in early stance [10] and more oscillations or rotational movements of the head, shoulders, and trunk [4]. These preliminary findings, if confirmed, would signify great functional disturbance in children with AS and may affect their activity participation [11]. Therefore, further research on the gait pattern in this particular group of children is deemed necessary.

The use of sound to improve health was first introduced by the famous ancient Greek philosophers Aristotle and Plato [12]. When compared to other kinds of external cues, auditory cues were found to be more matched with the rhythmical and continuous movement patterns of humans (e.g., walking) [13]. Nowadays, auditory cues are one of the most common treatment modalities used in neurological rehabilitation [14]. For example, patients with Parkinson's disease

demonstrate a smoother and more organized gait pattern under the influence of an external auditory cue [14,15], possibly because the sound cue improves attention and guides movements [16]. Children with AS, similar to patients with Parkinson's disease, also show structural deficits in the basal ganglia and have movement deficits [17-19]. Therefore, we hypothesized that rhythmical auditory stimulation (RAS)-assisted gait training might also improve the walking pattern in children with AS. This study had two aims: (1) to evaluate gait abnormalities in children with AS and (2) to investigate the immediate and carry-over effects of RAS-assisted gait training on the gait pattern in this particular group of children.

Materials and Methods

Participants

Children with AS were recruited from local Child Assessment Centres, non-government organizations, and the clinic of our University. Inclusion criteria were (1) a formal diagnosis of AS according to the DSM-IV-TR [2], (2) age of 6 to 12 years old, (3) no known co-morbidities, and (4) studying in mainstream schools. Exclusion criteria were (1) formal diagnosis of emotional, neurological, hearing, visual, movement, or other psychiatric disorders, (2) significant musculoskeletal injuries or cardiopulmonary conditions that may influence walking pattern, (3) children who could not follow simple

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instructions, (4) taking medication related to AS, or (5) participating regularly in sports activities. For the control group, age-matched children with normal development were recruited from the community on a voluntary basis using the same inclusion and exclusion criteria, except that they had no diagnosis of AS.

Procedures

The study was approved by the Human Subjects Ethics Review Subcommittee of the administering University, and all procedures were conducted in accordance with the Declaration of Helsinki. After explaining the study to each participant and their parents, written informed consent was obtained. The entire measurement process was carried out in the Motion Analysis Laboratory of our university. A VICON 370 system with eight cameras and computer software (Oxford Metrics Ltd., Oxford, United Kingdom) were used to capture the gait pattern during the three testing conditions (i.e., baseline, sound-on, and sound-off) (Figure 1) and to interpret the kinematic gait parameters afterward.

Each participant was instructed to wear a swimming suit during the test. Reflective markers were placed on the participant's sacrum (mid-way between the posterior superior iliac spines on both sides), bilateral anterior superior iliac spines, thighs (lower lateral one-third surface of the thigh), knee joints (lateral epicondyle of femur), lateral tibiae (lower one-third of the shank), ankle joints (lateral malleolus), calcaneus, and second metatarsal heads (Figure 2) according to the specifications of the VICON 370 system [20] by an experienced pediatric physiotherapist. The participant then walked barefoot on a treadmill at a comfortable speed for 1 minute as a familiarization trial. For the actual test, the participant walked on the treadmill for 5 minutes at his or her preferred walking speed with no auditory cue (i.e., the baseline testing condition) (Figure 1). The treadmill speed (mph) and cadence (steps/minute) were recorded. A 5-minute break was allowed after this baseline measurement.

The second testing condition was walking with an auditory cue (sound-on condition) (Figure 1). A digital dual metronome reference pitch generator (Intelli IMT-1000, New Market VA, United States) was used to generate the auditory cue. To standardize the step frequency across the testing conditions, the frequency of the metronome was adjusted to the average cadence adopted by the participant during the baseline measurement. The participant was then asked to walk

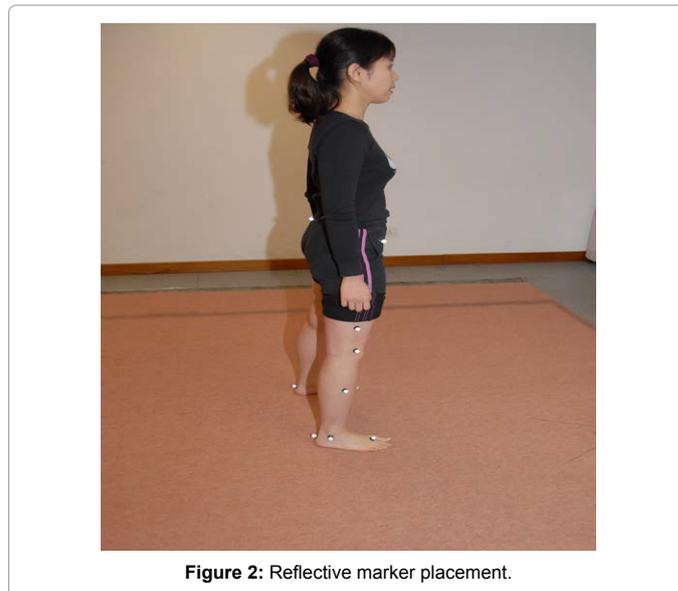


Figure 2: Reflective marker placement.

according to the pace of the metronome for 5 minutes without stopping. The treadmill speed was kept constant (i.e., same speed as used in the baseline measurement). Another 5-minute rest was given afterward (Figure 1).

The third testing condition was walking without the auditory cue (sound-off condition) (Figure 1). The participant was asked to walk on the treadmill at the same speed at a similar step frequency for 5 minutes, but no verbal or visual feedback was given. Outcome measures including average step length (anterior-posterior distance from heel-strike of one foot to toe-off of the contralateral foot), step width (medial-lateral distance between the locations of sequential left and right heel-strikes) [21], step height (maximum distance between the heel of the swing leg and the ground), total pelvic rotation angle, percentage of time spent in the double-leg supporting (DLS) phase, and hip and knee joint angles at mid-stance phase of gait [20] throughout the three testing conditions were used for analysis (Figure 1).

Statistical analysis

The demographic results are expressed as mean \pm standard deviation (SD). The Mann-Whitney U-test was used to compare the age, height, and weight between the two groups, whereas sex was compared with the Chi-square test. Comparison of gait parameters between the baseline, sound-on, and sound-off conditions was performed with the Friedman test for repeated measures followed by multiple Wilcoxon signed-rank tests if the result was statistically significant. Moreover, the Mann-Whitney U-test was used to compare all of the outcome gait measures between the two groups. A significance level of 0.05 was adopted for all statistical tests and a Bonferroni correction per group of outcome gait parameters and per condition (baseline, sound-on, and sound-off) was applied to reduce the likelihood of false-positive results, leading to a level of significance of 0.017. All statistical procedures were performed with SPSS version 17.0.

Results

The demographic results are listed in (Table 1), and individual participant characteristics and detailed gait variables are presented in (Table 2). No significant differences were found among the baseline, sound-on, and sound-off gait parameters in either group ($p > 0.017$)

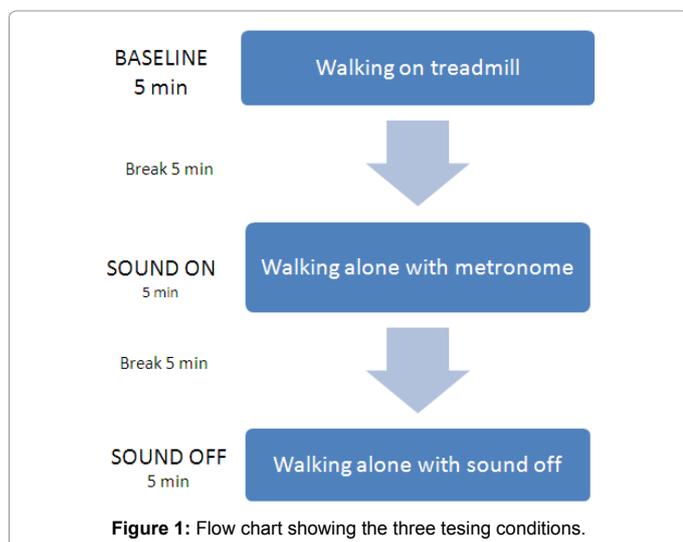


Figure 1: Flow chart showing the three testing conditions.

	Asperger group (n=5)	Control group (n=6)	p-value
Age (years)	8.5 ± 2.0	9.5 ± 2.0	0.712
Sex (n)	6 boys	3 boys, 2 girls	0.182
Height (cm)	90 ± 12.9	93.2 ± 8.0	0.776
Weight (kg)	21.0 ± 2.0	24.0 ± 3.0	0.314

Table 1: Demographic data of the participants (mean ± SD).

Age (years)	Sex	Group	Step length (cm)	Step width (cm)	Step height (cm)	Pelvic rotation angle (degrees)	% of time in DLS phase (%)	Hip joint angle at mid-stance (degrees)	Knee joint angle at mid-stance (degrees)
7.00	Male	Asperger	Baseline: 226.86 Sound-on: 228.06 Sound-off: 243.05	Baseline: 48.94 Sound-on: 47.28 Sound-off: 45.89	Baseline: 118.79 Sound-on: 118.22 Sound-off: 123.59	Baseline: 5.57 Sound-on: 5.45 Sound-off: 4.98	Baseline: 28.43 Sound-on: 27.92 Sound-off: 29.51	Baseline: 0.36 Sound-on: 0.29 Sound-off: -0.16	Baseline: 6.61 Sound-on: 6.37 Sound-off: 4.59
10.08	Male	Asperger	Baseline: 288.81 Sound-on: 290.72 Sound-off: 285.40	Baseline: 61.45 Sound-on: 66.89 Sound-off: 53.96	Baseline: 131.04 Sound-on: 129.98 Sound-off: 132.00	Baseline: 5.48 Sound-on: 5.55 Sound-off: 5.37	Baseline: 31.84 Sound-on: 31.37 Sound-off: 32.09	Baseline: -0.33 Sound-on: -0.35 Sound-off: -0.31	Baseline: 6.09 Sound-on: 6.02 Sound-off: 6.01
6.92	Male	Asperger	Baseline: 270.93 Sound-on: 271.87 Sound-off: 303.72	Baseline: 59.74 Sound-on: 57.60 Sound-off: 54.26	Baseline: 142.81 Sound-on: 141.25 Sound-off: 145.94	Baseline: 3.07 Sound-on: 3.50 Sound-off: 4.05	Baseline: 26.61 Sound-on: 28.77 Sound-off: 28.87	Baseline: -2.22 Sound-on: -2.12 Sound-off: -2.09	Baseline: -6.20 Sound-on: -6.03 Sound-off: -6.03
7.00	Male	Asperger	Baseline: 290.34 Sound-on: 297.55 Sound-off: 287.00	Baseline: 72.33 Sound-on: 42.17 Sound-off: 58.50	Baseline: 128.23 Sound-on: 138.70 Sound-off: 130.89	Baseline: 8.18 Sound-on: 5.29 Sound-off: 8.10	Baseline: 31.21 Sound-on: 30.29 Sound-off: 31.77	Baseline: 1.85 Sound-on: 1.95 Sound-off: 2.07	Baseline: 8.39 Sound-on: 7.72 Sound-off: 8.02
7.25	Male	Asperger	Baseline: 269.13 Sound-on: 268.43 Sound-off: 226.32	Baseline: 55.84 Sound-on: 51.07 Sound-off: 78.36	Baseline: 107.23 Sound-on: 137.75 Sound-off: 119.18	Baseline: 9.04 Sound-on: 9.18 Sound-off: 7.66	Baseline: 29.73 Sound-on: 28.86 Sound-off: 32.66	Baseline: 1.94 Sound-on: 1.80 Sound-off: 1.98	Baseline: 11.16 Sound-on: 11.89 Sound-off: 10.67
6.50	Male	Asperger	Baseline: 246.85 Sound-on: 248.91 Sound-off: 250.33	Baseline: 127.63 Sound-on: 111.20 Sound-off: 116.58	Baseline: 130.70 Sound-on: 134.45 Sound-off: 133.15	Baseline: 6.48 Sound-on: 6.09 Sound-off: 5.22	Baseline: 29.56 Sound-on: 28.96 Sound-off: 28.32	Baseline: 0.34 Sound-on: 0.44 Sound-off: 1.07	Baseline: 5.11 Sound-on: 5.21 Sound-off: 7.79
Asperger group mean ± SD: 7.46 ± 1.31	Boys: 6 Girls: 0	Asperger group mean ± SD:	Baseline: 265.49±24.67 Sound-on: 267.59 ± 25.93 Sound-off: 265.97 ± 30.29	Baseline: 70.99±28.79 Sound-on: 62.70 ± 25.26 Sound-off: 67.93 ± 26.20	Baseline: 126.47±12.15 Sound-on: 133.39 ± 8.39 Sound-off: 130.79 ± 9.20	Baseline: 6.30±2.13 Sound-on: 5.84 ± 1.86 Sound-off: 5.90 ± 1.61	Baseline: 29.56±1.89 Sound-on: 29.36 ± 1.24 Sound-off: 30.54 ± 1.85	Baseline: 0.32±1.54 Sound-on: 0.33 ± 1.50 Sound-off: 0.43 ± 1.60	Baseline: 5.19±5.97 Sound-on: 5.20 ± 5.99 Sound-off: 5.17 ± 5.86
6.50	Female	Control	Baseline: 430.65 Sound-on: 444.72 Sound-off: 437.43	Baseline: 41.36 Sound-on: 35.54 Sound-off: 40.31	Baseline: 161.58 Sound-on: 164.40 Sound-off: 163.49	Baseline: 2.30 Sound-on: 1.72 Sound-off: 2.24	Baseline: 23.09 Sound-on: 23.49 Sound-off: 23.15	Baseline: -2.16 Sound-on: -2.16 Sound-off: -2.41	Baseline: -13.36 Sound-on: -13.80 Sound-off: -16.34
12.00	Male	Control	Baseline: 427.50 Sound-on: 418.64 Sound-off: 415.67	Baseline: 46.74 Sound-on: 51.57 Sound-off: 50.25	Baseline: 170.90 Sound-on: 171.00 Sound-off: 167.96	Baseline: 4.26 Sound-on: 4.17 Sound-off: 4.14	Baseline: 24.77 Sound-on: 22.08 Sound-off: 23.39	Baseline: 2.09 Sound-on: 2.05 Sound-off: 2.07	Baseline: 7.99 Sound-on: 8.45 Sound-off: 10.34
9.00	Male	Control	Baseline: 453.87 Sound-on: 246.39 Sound-off: 500.51	Baseline: 83.91 Sound-on: 51.53 Sound-off: 80.83	Baseline: 199.69 Sound-on: 108.18 Sound-off: 209.57	Baseline: -0.03 Sound-on: 1.19 Sound-off: 0.77	Baseline: 20.68 Sound-on: 13.90 Sound-off: 18.98	Baseline: 1.04 Sound-on: 0.87 Sound-off: 1.02	Baseline: 1.92 Sound-on: 2.02 Sound-off: 1.88

7.50	Male	Control	Baseline: 179.20 Sound-on: 227.65 Sound-off: 251.14	Baseline: 37.40 Sound-on: 19.09 Sound-off: 26.64	Baseline: 111.35 Sound-on: 117.35 Sound-off: 120.12	Baseline: 5.32 Sound-on: 3.46 Sound-off: 5.56	Baseline: 32.60 Sound-on: 26.97 Sound-off: 27.01	Baseline: 0.87 Sound-on: 1.34 Sound-off: 1.53	Baseline: 8.36 Sound-on: 7.97 Sound-off: 7.82
6.50	Female	Control	Baseline: 373.48 Sound-on: 411.37 Sound-off: 393.34	Baseline: 47.85 Sound-on: 47.98 Sound-off: 46.91	Baseline: 151.71 Sound-on: 151.26 Sound-off: 152.13	Baseline: 1.71 Sound-on: 1.83 Sound-off: 1.74	Baseline: 22.53 Sound-on: 21.31 Sound-off: 21.07	Baseline: -1.93 Sound-on: -2.34 Sound-off: -2.24	Baseline: -5.80 Sound-on: -6.63 Sound-off: -4.86
Control group mean ± SD: 8.30 ± 2.31	Boys: 3 Girls: 2	Control group mean ± SD:	Baseline: 372.94 ± 112.24 Sound-on: 349.75 ± 103.87 Sound-off: 399.62 ± 92.13	Baseline: 51.45 ± 18.63 Sound-on: 41.14 ± 13.97 Sound-off: 48.99 ± 19.97	Baseline: 159.05 ± 32.12 Sound-on: 142.44 ± 28.19 Sound-off: 162.65 ± 32.21	Baseline: 2.71 ± 2.11 Sound-on: 2.48 ± 1.27 Sound-off: 2.89 ± 1.93	Baseline: 24.73 ± 4.64 Sound-on: 21.55 ± 4.79 Sound-off: 22.72 ± 2.99	Baseline: -0.02 ± 1.91 Sound-on: -0.05 ± 2.06 Sound-off: -0.01 ± 2.15	Baseline: -0.18 ± 9.35 Sound-on: -0.40 ± 9.65 Sound-off: -0.23 ± 10.75

Table 2: Detailed gait parameters of all participants.

	Baseline			Sound-on			Sound-off		
	Asperger group (n=6)	Control group (n=5)	Mann-Whitney U test p-value	Asperger group (n=6)	Control group (n=5)	Mann-Whitney U test p-value	Asperger group (n=6)	Control group (n=5)	Mann-Whitney U test p-value
Step length (cm)	265.49 ± 24.67	372.94 ± 112.24	0.100	267.59 ± 25.93	349.75 ± 103.87	0.465	265.97 ± 30.29	399.62 ± 92.13	0.028
Step width (cm)	70.99 ± 28.79	51.45 ± 18.63	0.068	62.70 ± 25.26	41.14 ± 13.97	0.201	67.93 ± 26.20	48.99 ± 19.97	0.144
Step height (cm)	126.47 ± 12.15	159.05 ± 32.12	0.068	133.39 ± 8.39	142.44 ± 28.19	0.584	130.79 ± 9.20	162.65 ± 32.21	0.068
Pelvic rotation angle (degrees)	6.30 ± 2.13	2.71 ± 2.11	0.018	5.84 ± 1.86	2.48 ± 1.27	0.011*	5.90 ± 1.61	2.89 ± 1.93	0.068
% of time in DLS phase (%)	29.56 ± 1.89	24.73 ± 4.64	0.100	29.36 ± 1.24	21.55 ± 4.79	0.006*	30.54 ± 1.85	22.72 ± 2.99	0.006*
Hip joint angle at mid-stance (degrees)	0.32 ± 1.54	-0.02 ± 1.91	0.855	0.33 ± 1.50	-0.05 ± 2.06	0.855	0.43 ± 1.60	-0.01 ± 2.15	0.715
Knee joint angle at mid-stance (degrees)	5.19 ± 5.97	-0.18 ± 9.35	0.361	5.20 ± 5.99	-0.40 ± 9.65	0.465	5.17 ± 5.86	-0.23 ± 10.75	0.465

*Bonferroni adjusted p-value < 0.017

Table 3: Gait parameters of the two groups (mean ± SD).

(Table 3). In the between-group comparisons, children with AS rotated their pelvis more during walking than did the control group at baseline ($p=0.018$) and during the sound-on ($p=0.011$) condition (Table 3). When the sound was off, there was no statistically significant difference in the pelvic rotation angle between the two groups ($p=0.068$) (Table 3).

The time spent in the DLS phase was also significantly different between the two groups in both the sound-on ($p=0.006$) and sound-off ($p=0.006$) conditions although there was no significant between-group difference at baseline. The control group spent less time in the DLS phase in the sound-on condition and the subsequent sound-off condition (Table 3). No statistically significant differences were found between the two groups in all other gait parameters (step length, step width, step height, and hip and knee joint angles at mid-stance phase of gait) in the three testing conditions (Table 3).

Discussion

Gait patterns in children with AS

Our results revealed that the children with AS had greater pelvic (trunk) rotational movements during walking on treadmill at comfortable walking speeds than did the children with normal development. Their step length, step width, step height, and hip and knee joint kinematics were all similar to those of the children with normal development. In addition, the time spent in the DLS phase, which is a reflection of gait stability, was also comparable between the two groups at baseline. These results are actually in line with several previous studies. For example, both Damasio and Maurer [22] and

Rinehart et al. [18] showed that children with AS had abnormal head and trunk movements during locomotion when compared to children with normal development. The excessive trunk rotation during walking might be explained by the poorer proprioceptive function [23], dysfunction of the dopaminergic system [24], and less distinct regional activation/deactivation of the basal ganglia [19] among children with AS. However, the reason for the movement deviation occurring only in the trunk/pelvis remains unknown. Further neuro-imaging and physiological studies are required to confirm the exact causes of excessive pelvic/trunk rotation during walking in children with AS.

Effect of RAS on gait pattern

Although there were no differences in gait parameters among the three testing conditions in both groups, comparison between groups did reveal a significant difference in the pelvic rotation angle when the sound cue was on. The control group had less pelvic rotation than the AS group when walking with RAS, but this effect was not maintained when the sound cue was switched off (Table 3). It seems that RAS was more useful in modifying the rotational gait pattern in typically developing children than those with AS. This finding actually further reinforces our postulation that the excessive pelvic (trunk) rotation during walking in the AS-affected children might be caused by structural deficits in the brain (e.g., dysfunction of the dopaminergic system and basal ganglia) [19,24] and poor proprioceptive function [23]. Therefore, their gait pattern might not be easily corrected by a short period of RAS-assisted gait training.

Apart from the lesser pelvic rotation, children in the control group

also had shorter DLS time than the AS group when the sound cue was on. This effect was carried over to the subsequent sound-off condition. Furthermore, the step length was longer in the control group than in the AS group when the sound was switched off (Table 2). All of these findings support the idea that RAS might be more effective in improving the gait pattern in children with normal development rather than in children with AS. This is in contrast to our original hypothesis that RAS-assisted gait training might improve the gait pattern in children with AS. These unexpected findings could be explained by our small sample size. Statistical power could be compromised, and so most of our findings would not be statistically significant. Moreover, the RAS-assisted gait training period of only 5 minutes might be too short to induce any significant effect on the walking pattern of the children with AS. Further studies should include a larger sample of participants and a longer duration of gait training with RAS.

Conclusion

Children with AS demonstrated excessive pelvic rotation during walking when compared to children with normal development. A short period of gait training with RAS might not be able to improve the walking pattern in children with AS.

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Declaration of Conflicting Interests

The authors declare that they have no conflicts of interest with respect to the authorship or publication of this paper. No funding was provided for its preparation.

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