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RESEARCH ARTICLE

Proprioception and vestibular alterations affect postural control in children with mild autism: A pilot study [version 1; peer review: 1 approved, 1 approved with reservations]

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Abstract
Background: Individuals diagnosed with Autism Spectrum Disorder (ASD) exhibit some type of motor control impairment, for instance, motor apraxia and history of gross motor delay that could lead to increased risk of fall. This pilot research was designed to assess and characterize static postural stability and create a starting point to better understand and describe postural control in children with mild autism.
Method: We measured static postural control with center of pressure (COP) displacement in 10 children with mild autism during eight sensory conditions that challenge and cancel the visual, proprioceptive and vestibular systems.
Results: Our results showed that children with autism demonstrated increased postural sway in response to challenges to the proprioceptive and vestibular systems.
Conclusion: Therefore, under appropriate, challenging conditions, static postural control instability can be detected in children with mild autism.

Keywords
Autism Spectrum Disorder, Static Postural Control, Sway

Open Peer Review

Approval Status ? ✓

	1	2
version 1 12 Mar 2018	? view	✓ view

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2. **Juan Carlos Galloza-Otero**, University of Texas Health Science Center at Houston, Houston, USA

Any reports and responses or comments on the article can be found at the end of the article.

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Author roles: Rosario MG: Conceptualization, Data Curation, Formal Analysis, Investigation, Methodology, Project Administration, Resources, Supervision, Validation, Visualization, Writing – Original Draft Preparation, Writing – Review & Editing; **López L**: Formal Analysis, Investigation, Methodology, Writing – Original Draft Preparation, Writing – Review & Editing; **Méndez M**: Investigation, Methodology, Writing – Original Draft Preparation, Writing – Review & Editing; **Ababneh AF**: Investigation, Writing – Original Draft Preparation, Writing – Review & Editing; **Gonzalez-Sola M**: Investigation, Writing – Original Draft Preparation, Writing – Review & Editing

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Introduction

Autism is a developmental disorder characterized by deficits in language, social skills, environment interaction and motor function development; although the latter has not been thoroughly researched (Mari *et al.*, 2003) (Fournier *et al.*, 2010a). According to the "Diagnostic and Statistical Manual of Mental Disorders" (DSM-IV), classic autism, as well as Asperger's disorder, infantile disintegrative disorder and generalized not specified developmental disorder is now all classified under the Autism Spectrum Disorder (ASD) (American Psychiatric Association, 2013).

Individuals diagnosed with Autism Spectrum Disorder (ASD) have been shown to have deficiencies in motor control such as motor apraxia, clumsiness, reduced ankle movement, history of gross motor delay, and toe-walking (Ming *et al.*, 2007; Teitelbaum *et al.*, 1998). These deficiencies could lead to a risk of falls and lower quality of life in these individuals.

Kohen-Raz *et al.* (1992) found that children with autism have more difficulty maintaining postural control due to the gait pattern (walk on tip-toes) that they exhibit regularly. Also, children with ASD have difficulty in the integration of sensory information. Specifically, deficits may exist in the processing of vestibular and proprioceptive information when compared to children with normal development (Blanche *et al.*, 2012; Fournier *et al.*, 2010b; Shumway-Cook & Woollacott, 2001a).

The literature shows concurrent limitations and consensus in which sensory system specifically alter postural control in children with autism. Nevertheless, problems in motor control may be caused by some deficit in postural control mechanisms (visual, proprioceptive and vestibular systems) or problems in the integration/adaptation of these systems (Blanche *et al.*, 2012; Fournier *et al.*, 2010a; Fournier *et al.*, 2010b; Kern *et al.*, 2007; Kohen-Raz *et al.*, 1992; Molloy *et al.*, 2003; Shumway-Cook & Woollacott, 2001b).

Consequently, it is necessary to study and describe how the sensory systems relate to postural control in children with ASD. The primary purpose of our research project is to identify postural deficiencies that may exist and to describe postural control in children who have autism with the objective to ascertain how these systems react when challenged or altered. Therefore, we hypothesized that when standing still (double legged), children with ASD will exhibit the following: 1) increased sway in one or more of the balance conditions, 2) challenges of the vestibular and proprioceptive systems will increase center of pressure (COP), and sway in a mediolateral (M-L) direction.

The long term goals of this pilot study are to provide the foundation to enhance the understanding of postural control, create new inquiry projects to help comprehend postural control in the different categories of autism and aid in developing targeted interventions for fall prevention in children with autism.

Materials and methods

Parents of children diagnosed with mild ASD who were interested in the study called the number on the recruitment flyer. The PI spoke with the parent to assess whether their child qualified for the study based on the following inclusion and exclusion criteria. The inclusion criteria were: (1) children from 7–12 years of age, male or female, (2) mild ASD diagnosis (as determined by a medical doctor (3) (5) capable of ambulating independently. Exclusion criteria were: (1) children with additional neurological problems, (2) children with visual problems, (3) children unable to tolerate walking or standing barefoot, (4) children who have fallen three or more times in the last three months, and (5) children with vestibular problems. We assessed the vestibular system utilizing the Fukuda Stepping Test (Fukuda, 1959). After hearing the qualifications, if the parent wished to volunteer their child for the study, an appointment was made for them to come to the Biomechanical Laboratory.

Ten children, diagnosed with a mild ASD that fulfilled the inclusion criteria participated in this study (Table 1). This project was conducted in the Biomechanical Laboratory of the Medical Science Campus of the University of Puerto Rico. The recruitment of participants was performed by posting flyers at the University of Puerto Rico, Medical Science Campus and other Centers in the Metropolitan Area.

After parents and children had given written consent, the following steps were followed: (1) A preparatory protocol was performed to familiarize the children with the study protocol, the staff and the children spent 5 minutes watching photos related to the materials, equipment, workplace, and team members. (2) Anthropometric measurements were taken (weight and height) as descriptive measures and to calibrate the pressure mat, MatScan. (3) The Fukuda test was performed to rule out vestibular system impairment (Fukuda, 1959).

Balance assessment protocol

To assess balance, we used a MatScan TM pressure mat (TekScan, Boston, MA). This mat contains sensors that measure body sway in centimeters (cm), anteriorly (forward), posteriorly (backward) (A–P), or laterally (sideways) (L–R). The mat provides information about the direction and amount of sway as well

Table 1. Demographic and Anthropometric Information of Participants.

Gender		*Mean ± SD*		
		Weight (kg)	**Age (years)**	**Height (m)**
	M = 8	38.26 ± 6.12	8.9 ± 1.36	1.41 ± 0.08
	F = 2	22.5 ± 4.11	7.0 ± 0.00	1.26 ± 0.09
Total	10	35.11 ± 8.67	8.5 ± 1.43	1.38 ± 0.10

kg: kilogram; **m:** meter; **SD:** standard deviation as area or center of pressure (COP). The data collected with the pressure mat, center of pressure (cm^2) and sway (cm), was analyzed with Tekscan Sway Analysis Module (SAM) software designed for this purpose. This test was used to determine the effectiveness of an individual's ability to use different sensory stimuli to examine the balancing of the body while standing under different conditions. The MatScan has an intra-rater reliability of .96-1.0 (Zammit *et al.*, 2010).

The balance assessment procedure included the following; each subject was instructed to stand on both feet for 15 seconds on the pressure MatScan under eight different conditions. The first four conditions were executed over a stable surface and consisted of the following: (1) eyes open (EO) -evaluates visual, vestibular and proprioceptive systems, (2) eyes closed (EC) -eliminates visual input, evaluates vestibular and proprioceptive system, (3) eyes open while moving head up and down (at 60 beats per minute) (EO HUD) -evaluates visual and proprioceptive system and alters vestibular input, (4) eyes closed while moving the head up and down (EC HUD) -evaluates the effect of removing visual information, and the vestibular input being altered. The subjects then performed the same four tasks, this time standing on an unstable platform (high quality closed cell foam 19 inches long x 15 inches width x 2.25 inches) with the purpose of altering the proprioceptive input and increasing dependence on the visual and vestibular systems. (1) eyes open (EO MAT) -evaluates visual and vestibular system while the proprioceptive system is challenged and, (2) eyes closed (EC MAT) -removes visual input, evaluates the vestibular system and alters the proprioceptive system. (3) eyes open while moving the head up and down (EO MAT HUD) -evaluates visual system modifying the input of proprioceptive and vestibular systems, (4) eyes closed while moving the head up and down (EC MAT HUD) -evaluates altered proprioceptive and, vestibular systems in the absence of visual system.

Data analysis

The software used to analyze all the information was the statistical package for the social science version 19 (SPSS). P values <.05 were accepted as statistically significant. This study's focus was to identify postural control deficiency in children diagnosed with mild ASD. We assessed how participants reacted when exposed to a stable and unstable surface and how altered proprioceptive information impacts postural control. We used a ANOVA analyses to compare differences in COP and sways (AP and ML) within the eight balance sensory conditions. A Bonferroni post hoc analysis was used to identify the specific variables or parameters to which significant differences could be attributed.

Results

We recruited ten children diagnosed with mild ASD, eight male and two female with an age average of 8.58.5± 1.433 (anthropometric information are in Table 1).

We compared postural control between firm (stable) and foam (unstable) surface to identify postural instability. In detail, we used eye open on a firm surface test as a baseline and compared to the other three (eyes closed, eyes open head up and down, and eyes closed head up and down) conditions. Likewise, we examined eyes open on foam (unstable) to the other three conditions on the foam.

Results of the comparison of average COP and sway movement under four different conditions (EO, EC, EO HUD, and HUD EC) on a stable surface and the mean excursion of the COP and sways under the same four conditions on an unstable surface are shown in Table 2. In our study, the most significant results were that children showed significantly more oscillations (instability) on the comparison of unstable surface versus the stable condition.

Table 2. Mean and Standard Deviation of COP and AP & ML sways of the Stable and Unstable Surfaces Comparisons.

TESTS	Mean ± SD					
Stable Surface	**COP (cm²)**		**AP (cm)**		**ML (cm)**	
	EO (6.30 ± 5.63) vs. EC (4.86 ± 3.35)	*p* = .44	EO (5.70 ± 1.94) vs. EC (4.38 ± 2.03)	*p* = .55	EO (4.40 ± 2.00) vs. EC (2.73 ± 1.26)	*p* = .07
	EO (6.30 ± 5.63) vs. EOHUD (13.07 ± 7. 19)	*p* = .10	EO (5.70 ± 1.94) vs. EOHUD (6.21 ± 2.97)	*p* = .82	EO (4.40 ± 2.00) vs. EO HUD (4.88 ± 3.21)	*p* = .79
	EO (6.30 ± 5.63) vs. EC HUD (16.23 ± 13.69)	*p* = .17	EO (5.70 ± 1.94) vs. EC HUD (6.59 ± 2.42)	*P* = .67	EO (4.40 ± 2.00) vs. EC HUD (3.96 ± 2.01)	*p* = .79
Unstable Surface	**COP (cm²)**		**AP (cm)**		**ML (cm)**	
	EO MAT (41.72 ± 39.42) vs. EC MAT (39.84 ± 38.10)	p = .63	EO MAT (6.29 ± 4.50) vs. EC MAT (4.94 ± 3.38)	p = .48	EO MAT (11.04 ± 5.52) vs. EC MAT (11.84 ± 3.60)	*p* = .07
	EO MAT (41.72 ± 39.42) vs. EO MAT HUD(102.41 ± 79.12)	*p < .05*	EO MAT (6.29 ± 4.50) vs. EO MAT HUD(11.25 ± 5.88)	p = .06	EO MAT (11.04 ± 5.52) vs. EO MAT HUD(14.85 ± 5.38)	*p* = .79
	EO MAT (41.72 ± 39.42) vs. EC MAT HUD (99.01 ± 55. 32)	*p < .05*	EO MAT (6.29 ± 4.50) vs. EC MAT HUD (12.11 ± 4.77)	*P < .05*	EO MAT (11.04 ± 5.52) vs. EC MAT HUD (14.76 ± 5.42)	*p* = .79

COP: Center of Pressure; **EO:** Eyes Open; **EC:** Eyes Close; **HUD:** Head Up and Down; **MAT:** Pressure Mat; **AP:** antero-posterior; **ML:** medio-lateral; **SD:** standard deviation; **cm:** centimeters

Dataset 1. Standing Postural Control Pressure Data

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Pressure data collected was CoP, center of Pressure displacement in centimeters squared and sways. Antero-posterior (AP) and laterally sideways data was measured in centimeters. All measurements were collected during the 8 sensory balance conditions. Eyes open, eyes closed, eyes open head movements and eyes closed head movements. Same as the previous test were analyzed, however on standing on a foam/unstable surface.

Discussion

The purpose of our study was to identify postural control deficiency, compare how participants react when exposed to a stable and unstable surface, and to describe how each system responds when altered. We identified postural instability and the systems that might be responsible for these alterations in these children with mild autism. Therefore, we discussed our findings in the following working hypothesis.

Hypothesis: children with ASD will exhibit altered postural instability in one or more of the balance conditions. Under stable and unstable conditions, we identified postural instability on unstable condition. Similar to Molloy and colleagues (2003) we found that children with ASD exhibited increased oscillations when standing on the foam, or unstable surface, compared to a stable platform, thus showing postural alterations (Molloy *et al.*, 2003). Therefore, our hypothesis is accepted.

Participants showed increased COP excursion (postural instability) on the stable surface under EC HUD condition (when the proprioceptive is the only unaltered system, however not significant at this point. It is possible that the proprioceptive system could not compensate correctly when the input of the visual was canceled, and when altering the vestibular input, to maintain postural control.

During stable surface under all conditions, AP oscillations predominated. Usually, this is expected because human oscillations occur in AP axis due to knee and ankle joints, similar to an inverted pendulum, thus allowing slight medio-lateral movements (Shumway-Cook & Woollacott, 2001b). However, when these oscillations exceed the stability limits, it might be due to weakness in the muscles of the lower extremities or delay of the integration or adaptation of the proprioceptive system (Shumway-Cook & Woollacott, 2001b).

Postural control on an unstable surface shows that our participants exhibit greater COP excursion during the EO MAT HUD and EC MAT HUD condition. This condition was designed to evaluate the role of the visual system input. Therefore, we believe that the visual system was not capable helping to maintain proper balance when the vestibular and proprioceptive were stimulated. However, we believe that the visual system was not affected; it is just not enough to compensate for the other two tested systems. While the participants moved their heads up and down (in a coordinated fashion), they were requested to gaze at a fixed point during the entire task. Because of their low capability of concentration, it was not possible for these children to stare at a fixed point during the whole task. Our participants (when challenging the proprioceptive system) exhibited a trend of movements to the right and anterior directions showing further instability in each one of the tests. Similarly, when altering the visual and vestibular inputs, it was observed a tendency to move anterior and to the right. These results lead us to believe that proprioceptive input in children with autism could interfere with the ability of this system to send the correct information to maintain proper postural stability. (Blanche *et al.*, 2012; Fournier *et al.*, 2010b; Hansson *et al.*, 2010; Shumway-Cook & Woollacott, 2001b). Contrary to a stable surface, ML oscillations predominated under all conditions on the unstable surface (Table 2). One possible explanation for this event may be that the alteration of the proprioceptive system is responsible for the adjustment of the activation of the musculature of the trunk, thus causing instability in the ML direction (Blanche *et al.*, 2012; Fournier *et al.*, 2010b; Hansson *et al.*, 2010). Another possible justification might be related to balance control in the ML direction, mainly occurring due to weakness at the hip and trunk areas. Therefore, we believe that weakness in the trunk and hip muscles and deficits in weight bearing distribution might cause an increase in ML oscillations (O'sullivan, 2007; Shumway-Cook & Woollacott, 2001b).

In conclusion, children with mild autism spectrum disorder showed postural instability on the stable and unstable surfaces. Our study established that these children exhibited an increase in oscillations both for the AP and ML directions; however, the direction varied depending on what systems were altered. Thus, under controlled test conditions, children with autism exhibit diminished postural control or postural instability due to vestibular and proprioceptive alteration and postural instability in an ML direction.

Data availability

Standing postural control pressure data

Pressure data collected was CoP, center of Pressure displacement in centimeters squared and sways. Antero-posterior (AP) and laterally sideways data was measured in centimeters. All measurements were collected during the 8 sensory balance conditions. Eyes open, eyes closed, eyes open head movements and eyes closed head movements. Same as the previous test were analyzed, however on standing on a foam/unstable surface. 10.5256/f1000research.14179.d197064 (Rosario *et al.*, 2018)

Ethics and consent

Institutional Review Board (IRB) approval at the University of Puerto Rico, Medical Science Campus was obtained prior the recruitment of the participants. We obtained informed consent from all individuals (children and parents) included in the investigation.

Competing interests

The authors have no competing interests to disclose.

Grant information

The author(s) declared that no grants were involved in supporting this work.

Acknowledgments

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Open Peer Review

Current Peer Review Status: ? ✓

Version 1

Reviewer Report 15 May 2018

https://doi.org/10.5256/f1000research.15424.r31806

✓ **Juan Carlos Galloza-Otero**
Department of Physical Medicine and Rehabilitation, McGovern Medical School, TIRR Memorial Hermann, University of Texas Health Science Center at Houston, Houston, TX, USA

I understand that this is a pilot study and that the task of recruiting patients with the appropriate inclusion criteria may be very difficult. Regarding the study design, it would have been valuable to have an age-matched control group similar as it was done on the referenced study by Fournier *et. al*.[1] They studied the postural control of 13 children with Autism Spectrum Disorders and had 12 age-matched controls. Even though the sample was also small, having a control group in this study added more value to the findings and final conclusions. In the reviewed study by Rosario *et. al.* the absence of a control group in the study design should be mentioned as a limitation that may hinder the conclusions drawn from the results. This being said, there is significant scientific value to this study and more data is needed to evaluate potential postural control issues in patients with autism spectrum disorders.

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Is the work clearly and accurately presented and does it cite the current literature?
Yes

Is the study design appropriate and is the work technically sound?
Partly

Are sufficient details of methods and analysis provided to allow replication by others?
Yes

If applicable, is the statistical analysis and its interpretation appropriate?
I cannot comment. A qualified statistician is required.

Are all the source data underlying the results available to ensure full reproducibility?
Yes

Are the conclusions drawn adequately supported by the results?
Partly

Competing Interests: No competing interests were disclosed.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Reviewer Report 09 April 2018

https://doi.org/10.5256/f1000research.15424.r32378

?

Amit Bhattacharya
University of Cincinnati , Cincinnati, OH, USA

This pilot study with 10 subjects (8 males and 2 females) provided assessment of postural balance in children with mild autism using a Matscan pressure mat. Since the purpose of pilot study is to keep minimal variability among subjects' demographics, what was the rationale of having only two female subjects? This small sample size of female subjects needs to be justified.

Since COP measured with MatScan pressure mat sensors provides area (cm*cm) of pressure distribution under the sole, the results cannot be comparable to sway area measured by the "gold standard" force plate which is a measure of area contained within x-y distribution of center of pressure sway positions of whole body during the balance test. The discussion section needs to address the issues of concerns about the differences in measures of COP with a MatScan pressure mat vs with a Force Plate. Recent 2018 article by Goetschius *et. al*[1] reports that the pressure-mat COP measurements are smaller than those of the force-plate, and the differences between devices appeared to increase as the measurement magnitude increased. Goetschius et. al (2018) also reported that data collection period longer than 10 sec (15 sec test was used in the current manuscript) may demonstrate an even greater discrepancy in time dependent metrics between a MatScan pressure mat and Force plate devices. In other words, while a MatScan pressure mat data may be correlatable to those reported with a Force platform, the absolute values of postural balance outputs cannot be compared between two measurement systems. Therefore, measurements by a MatScan pressure mat and Force Plate should not be used interchangeably. Is it possible that under the feet pressure distribution pattern (instead or in addition to COP) captured by a MatScan pressure mat provides a unique signature of discrimination between mild autism and advanced autism? These issues need to be added as a limitations, as well as possible future utility of MatScan pressure mat metrics for quantifying proprioception feedback by the autism associated changes in pressure-receptors distribution patterns under the sole of feet.

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Is the work clearly and accurately presented and does it cite the current literature?
Partly

Is the study design appropriate and is the work technically sound?
Partly

Are sufficient details of methods and analysis provided to allow replication by others?
Yes

If applicable, is the statistical analysis and its interpretation appropriate?
I cannot comment. A qualified statistician is required.

Are all the source data underlying the results available to ensure full reproducibility?
Yes

Are the conclusions drawn adequately supported by the results?
Partly

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Biomechanics, Quantitative posturagraphy Gait assessment, bone fragility quantification, effect of low level toxic chemicals on motor functions

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.