Proprioception and vestibular alterations affect postural control in children with mild autism: A pilot study [version 1; referees: 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
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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

Competing interests: No competing interests were disclosed.

How to cite this article: Rosario MG, López L, Méndez M et al. Proprioception and vestibular alterations affect postural control in children with mild autism: A pilot study [version 1; referees: 1 approved, 1 approved with reservations] F1000Research 2018, 7:305 (doi: 10.12688/f1000research.14179.1)

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

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) 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.

<table>
<thead>
<tr>
<th>Gender</th>
<th>Mean ± SD</th>
<th>Age (years)</th>
<th>Height (m)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Weight (kg)</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>M = 8</td>
<td>38.26 ± 6.12</td>
<td>8.9 ± 1.36</td>
</tr>
<tr>
<td></td>
<td>F = 2</td>
<td>22.5 ± 4.11</td>
<td>7.0 ± 0.00</td>
</tr>
<tr>
<td></td>
<td>Total</td>
<td>35.11 ± 8.67</td>
<td>8.5 ± 1.43</td>
</tr>
</tbody>
</table>

kg: kilogram; m: meter; SD: standard deviation
as area or center of pressure (COP). The data collected with the pressure mat, center of pressure (cm²) 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.

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

<table>
<thead>
<tr>
<th>TESTS</th>
<th>Mean ± SD</th>
<th>COP (cm²)</th>
<th>AP (cm)</th>
<th>ML (cm)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Stable Surface</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>EO (6.30 ± 5.63) vs.</td>
<td>p = .44</td>
<td>EO (5.70 ± 1.94) vs.</td>
<td>p = .55</td>
<td>EO (4.40 ± 2.00) vs.</td>
</tr>
<tr>
<td>EC (4.86 ± 3.35)</td>
<td></td>
<td>EC (4.38 ± 2.03)</td>
<td></td>
<td>EC (2.73 ± 1.26)</td>
</tr>
<tr>
<td>EO (6.30 ± 5.63) vs.</td>
<td>p = .10</td>
<td>EO (5.70 ± 1.94) vs.</td>
<td>p = .82</td>
<td>EO (4.40 ± 2.00) vs.</td>
</tr>
<tr>
<td>EOHUD (13.07 ± 7.19)</td>
<td></td>
<td>EOHUD (6.21 ± 2.97)</td>
<td></td>
<td>EOHUD (4.88 ± 3.21)</td>
</tr>
<tr>
<td>EO (6.30 ± 5.63) vs.</td>
<td>p = .17</td>
<td>EO (5.70 ± 1.94) vs.</td>
<td>P = .67</td>
<td>EO (4.40 ± 2.00) vs.</td>
</tr>
<tr>
<td>EC HUC (16.23 ± 13.69)</td>
<td></td>
<td>EC HUC (6.59 ± 2.42)</td>
<td></td>
<td>EC HUC (3.96 ± 2.01)</td>
</tr>
<tr>
<td><strong>Unstable Surface</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>EO MAT (41.72 ± 39.42) vs.</td>
<td>p = .63</td>
<td>EO MAT (6.29 ± 4.50) vs.</td>
<td>p = .48</td>
<td>EO MAT (11.04 ± 5.52) vs.</td>
</tr>
<tr>
<td>EC MAT (39.84 ± 38.10)</td>
<td></td>
<td>EC MAT (4.94 ± 3.38)</td>
<td></td>
<td>EC MAT (11.84 ± 3.60)</td>
</tr>
<tr>
<td>EO MAT (41.72 ± 39.42) vs.</td>
<td>p &lt; .05</td>
<td>EO MAT (6.29 ± 4.50) vs.</td>
<td>p = .06</td>
<td>EO MAT (11.04 ± 5.52) vs.</td>
</tr>
<tr>
<td>EOHUD (102.41 ± 79.12)</td>
<td></td>
<td>EOHUD (11.25 ± 5.88)</td>
<td></td>
<td>EOHUD (14.85 ± 5.38)</td>
</tr>
<tr>
<td>EO MAT (41.72 ± 39.42) vs.</td>
<td>p &lt; .05</td>
<td>EO MAT (6.29 ± 4.50) vs.</td>
<td>P &lt; .05</td>
<td>EO MAT (11.04 ± 5.52) vs.</td>
</tr>
<tr>
<td>EC MAT HUC (99.01 ± 55.32)</td>
<td></td>
<td>EC MAT HUC (12.11 ± 4.77)</td>
<td></td>
<td>EC MAT HUC (14.76 ± 5.42)</td>
</tr>
</tbody>
</table>

**SD:** standard deviation; **cm:** centimeters

**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 an 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 ± 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 HUC 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.
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 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.

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**Dataset 1. Standing Postural Control Pressure Data**

http://dx.doi.org/10.5256/f1000research.14179.d197064

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.
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
Thanks to the University of Puerto Rico Medical Science Campus for providing the Biomechanical Laboratory and the MatScan equipment necessary for the development and completion of this study. Thanks to SER de Puerto Rico in San Juan, Puerto Rico for allowing us to recruit subjects in their facilities. This article was published with support from Texas Woman’s University Libraries’ Open Access Fund.

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References Source


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Open Peer Review

Current Referee Status:  ✔

Version 1

Referee Report 15 May 2018

doi: 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. 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.

References

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

Referee Report 09 April 2018

doi: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 (cm²) 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 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.

References

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.

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

I have read this submission. I 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.

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