Title: Postural mechanisms in moderate-to-severe cerebral palsy Running Title: Postural mechanisms in moderate-to-severe cerebral palsy **Authors**: Adam Goodworth^{1,2} & **Sandra Saavedra² ** Both authors contributed equally Author Affiliations where the work was done: 1. Department of Kinesiology, Westmont University, Santa Barbara, CA 2. Department of Rehabilitation Sciences, University of Hartford, 200 Bloomfield Avenue, West Hartford, CT 06117, USA Conflict of Interest: None **Acknowledgements:** We thank Yen-Hsun Wu for contribution to data collection and Cameron Abbot and Gabrielle Ruban for contribution to data analysis. Funding for this study was received from the National Science Foundation (DARE #1803714) awarded to A.D. Goodworth and S. Saavedra and NIH R03 DC013858 awarded to S. Saavedra and A.D. Goodworth. Corresponding Author: Adam Goodworth, agoodworth@westmont.edu

Abstract

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People with moderate-to-severe cerebral palsy (CP) have the greatest need for postural control research yet are usually excluded from research due to deficits in sitting ability. We use a support system that allows us to quantify and model postural mechanisms in non-ambulatory children with CP. A continuous external bench tilt stimulus was used to evoke trunk postural responses in 7 sitting children with CP (ages 2.5 to 13 years) in several test sessions. Eight healthy adults were also included. Postural sway was analyzed with root mean square (RMS) sway and RMS sway velocity, along with frequency response functions (FRF, gain and phase) and coherence functions across two different stimulus amplitudes. A feedback model (including sensorimotor noise, Passive, Reflexive, and Sensory Integration mechanisms) was developed to hypothesize how postural control mechanisms are organized and function. Experimental 35 results showed large RMS sway, FRF gains, and variability compared to adults. Modeling suggested that many subjects with CP adopted "simple" control with major contributions from a Passive and Reflexive mechanism and only a small contribution from active sensory integration. In contrast, mature trunk postural control includes major contributions from sensory integration and sensory reweighting. Relative to their body size, subjects with CP showed significantly lower damping, 3-5 times larger corrective torque, and much higher sensorimotor noise 40 compared to the healthy mature system. Results are the first characterization of trunk postural responses and the first attempt at system identification in moderate-to-severe CP, an important step toward developing and evaluating more targeted interventions.

News and Noteworthy

Cerebral palsy (CP) is the most common cause of motor disability in children. People with moderate-to-severe CP are typically non-ambulatory and have major impairments in trunk postural control. We present the first systems identification study to investigate postural responses to external stimulus in this population and hypothesize at how the atypical postural control system functions with use of a feedback model. People with moderate-to-severe CP may use a simple control system with significant sensorimotor noise.

Introduction

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- 55 Postural control of the trunk is a foundational motor skill, with the human trunk (head, arms,
- torso, and pelvis) comprising over half of the body's mass(De Leva 1996; Plagenhoef et al.
- 57 1983). Impaired trunk posture is a hallmark of severe cerebral palsy (CP). This impairment limits
- 58 nearly all voluntary activities, including speech, swallowing, eating(Redstone and West 2004;
- 59 Stevenson 1995), reaching(Santamaria et al. 2016; van der Heide et al. 2004), and
- 60 gait(Heyrman et al. 2014). People with severe CP present with many challenging clinical
- 61 features: unable to sit independently, abnormal muscle tone, dependent on wheelchair and
- caregivers for mobility, and are often non-verbal. These challenges have greatly limited postural
- research in this population. The purpose of this study is to better understand how moderate-to-
- severe CP impacts the trunk postural control system and to develop a hypothesis about how
- control mechanisms are diminished or intact using feedback modeling.
- 66 CP is the most common cause of motor disability in children: incidence of 2.5-3.6/1000 live
- births in the U.S.A.(CDC statistics from 2008 https://www.cdc.gov/ncbddd/cp/data.html).
- 68 Studies show motor development plateaus for children with CP as early as 3 years of age with 1
- out of 3 children unable to gain adequate trunk control for stable, independent sitting(Beckung et al.
- 70 2007; Rosenbaum et al. 2002). Learning trunk postural control early in life is a key indicator of future
- ambulation(Novak et al. 2012; Rosenbaum et al. 2002). Children with CP who exhibit poor trunk
- 72 control would typically be categorized in the Gross Motor Function Classification System as level III-
- 73 V, and we will refer to this group as moderate-to-severe. There is currently little evidence for
- 74 effective intervention for postural deficits in children with moderate-to-severe cerebral
- palsy(Fowler et al. 2007; Novak et al. 2013). Deficits in trunk control cascade into altered
- function and limited weight bearing mobility, leading to other health issues such as
- 77 osteoporosis, boney abnormalities and muscle weakness(Henderson et al. 2005; Stevenson et
- 78 al. 2006), ultimately worsening prognoses and increasing financial commitments. There are
- 79 increased efforts internationally for earlier diagnosis and increased intensity of treatment for
- 80 children with CP(McIntyre et al. 2011).
- 81 However, it is difficult to create optimal interventions without an understanding of the postural
- system in moderate-to-severe CP. To date, very few postural studies have been performed in
- this population. Historically, trunk postural control was viewed as an "all or nothing" (either one
- 84 could sit independent or not). People lacking ability for independent sitting were typically
- 85 excluded from postural control research. Butler and Saavedra were the first to popularize the
- notion that "partial" trunk control is possible(Butler et al. 2010). Specifically, if external trunk
- 87 support was provided around the pelvis and/or torso, people with moderate-to-severe CP could
- demonstrate trunk stability in all superior segments against gravity and external perturbations. In
- a separate study, the postural kinematics were investigated in children with severe CP during
- 90 quiet sitting on a stationary bench(Saavedra and Woollacott 2015). Children with severe CP had
- 91 large postural sway and difficulty aligning and stabilizing their head. This study represented a
- major step forward by documenting the feasibility of collecting kinematic postural data and
- 93 detailing the influence trunk support.
- 94 But limited information about the underlying control system can be obtained from guiet sitting
- alone(Boonstra et al. 2013; Pasma et al. 2014; van der Kooij et al. 2005). Mechanisms
- 96 underlying trunk posture include complex interactions due to its closed loop nature, making it
- 97 difficult to understand causal relations across body sway, neural control, and biomechanics.
- 98 Body sway impacts sensory feedback, sensory feedback impacts body sway, and multi-

- 99 segment biomechanics of the body (mass of segments, inertias, and interaction torques) have
- 100 frequency dependent effects on sway. The best method to overcome this challenge is to relate
- an external stimulus (eg, visual tilt, galvanic vestibular stimulation, push, surface motion, etc.) to
- body sway response. (Blouin et al. 2007; Fitzpatrick et al. 1996; Goodworth and Peterka 2009;
- Moorhouse and Granata 2007; Oie et al. 2002; van der Kooij et al. 2005; Welch and Ting 2008).
- The present study is the first to apply stimulus-response methods to investigate postural control
- mechanisms in CP. Control mechanisms have been summarized through distinct processes:
- reliance on sensory systems, integration of multisensory cues, sensory-to-motor
- transformations, joint stiffness and damping, and adaptations of mechanisms to optimize
- control(Ebenbichler et al. 2001; Horak and Macpherson 1996; Horak and Nashner 1986; Kuo
- 2005; Shumway-Cook and Woollacott 2012). Sensory-to-motor responses must be tightly
- controlled to avoid an unstable system because 1) gravity tends to accelerate segments away
- 111 from upright, 2) sensory integration has an inherent time delay, and 3) sensory feedback is
- associated with uncertainty(Faisal et al. 2008; Kuo 2005; Peterka 2002; van der Kooij and
- 113 Peterka 2011). In adults, control of trunk posture can be influenced by joint stiffness, reflexes,
- and sensory feedback from visual, vestibular, and somatosensory systems to balance body
- segments against gravity and respond to internal and external torques (Andreopoulou et al.
- 2015; Goodworth and Peterka 2009; Maaswinkel et al. 2015; van Dieen et al. 2018; Wu et al.
- 2016). Moreover, these mechanisms are adaptable to voluntary control (van Drunen et al.
- 118 2016), and the environmental context (Andreopoulou et al. 2015; Goodworth and Peterka 2009;
- 119 Maaswinkel et al. 2015). It is unknown how damage to the brain in CP impacts these postural
- 120 control mechanisms.
- Our first goal is to characterize postural responses in moderate-to-severe CP. In addition to time
- domain metrics, we also employ frequency response functions (FRFs) and coherence functions
- that detail the relation between external stimulus and body sway response. These frequency-
- based analyses provide a non-parametric representation of the postural system. Second, we
- distinguish postural response (the average response to a repeated external stimulus) from
- postural variability (the variability in response to a repeated stimulus). This is important because
- severe CP is associated with large postural sway but the underlying reasons and details are not
- known. Finally, we develop a closed loop feedback model of the postural control system using
- parametric representations of the various control mechanisms. In all our goals, we present
- individual results whenever feasible alongside group averages because CP is a heterogeneous
- 131 condition.
- Taken together, this is the first detailed investigation of postural control mechanisms in
- moderate-to-severe CP. Since trunk postural control is the most critical constraint to function in
- children who are non-ambulatory, we felt it was essential to investigate the control processes for
- this population of children. Many of the methods presented here may be applicable to other
- populations with impaired trunk postural control.

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Methods

- 139 Human subjects & test sessions
- All testing was completed under an approved IRB protocol with the University of Hartford.

Seven children with CP were included in this study. Children were tested across several sessions. For various personal and logistical reasons, data in this study come from 2 sessions with some children, and 3 or more sessions with other children. Across all subjects with CP, there were 21 total sessions included in this study. Each session was at least 2 and less than 6 months apart for each child. Table 1 shows each subject's age ranges from first to last test session, along with other key descriptors.

The GMFCS is a clinical scale that classifies across 5 different levels of severity based on gross motor skills, primarily posture and mobility(Palisano et al. 1997; Palisano et al. 2008). People with ability to walk independently are classified as level I or II while people who require assistive devices for walking are classified as level III and those who need support for sitting are classified as level IV or V. The Segmental Assessment of Trunk Control (SATCo) is a clinical test that provides greater detail into one's ability to maintain postural control over their trunk segments(Butler et al. 2010). In this test, subjects are provided external trunk support and then are assessed in their ability to demonstrate static posture (stay vertical for 20 s), reactive control (quickly return to vertical after a slight push on the trunk), and anticipatory trunk control (remain vertical while turning one's head right and left). Generally, subjects with more severe CP can only demonstrate static, reactive, and anticipatory control when provided with external support on the rib cage or under the axillae.

Table 1. Anthropometric and clinical test summary for subjects with cerebral palsy.

subject	Age (months)	Height (cm)	Weight (kg)	GMFCS level	SATCo
CP10	128-140	136-139	36-42	III	LT to UL
CP11	52-64	99-110	14-16	IV/V	MT
CP13	128-160	122-135	23-32	IV	UT to MT
CP15	52-57	109-110	16-16.2	V	MT
CP20	55-70	99-99	14-17	V	MT
CP23	25-43	88-104	12-18	IV	UT to LT
CP24	24-39	99-98	15-16	IV	MT to LT

Note, each subject with CP was tested across multiple sessions separated by 2 - 6 months. Anthropometric and clinical test ranges are shown from the first to last session. GMFCS was measured and confirmed during each session. The SATCo lists the most inferior level of external support where subjects demonstrated static, active, and reactive control. Four subjects changed their SATCo scores from the first to last test session. UT = upper-thoracic; MT = mid-thoracic; LT = lower-thoracic; UL = upper lumbar

Eight adults were also included in this study (mean age 28 ± 6 yrs SD, mean height 167 ± 7.1 cm SD, and mean mass 63.8 +/- 10 kg SD) and were tested in one session. While adults are obviously not age matched, they do provide a valuable benchmark of healthy mature postural control. We are careful to only interpret adult data as a benchmark and not imply an age-specific comparison. Future studies are planned to compare results with age matched and developmentally matched typically developing children (see Limitations section for more details).

Protocol in each session

Testing postural control in children with moderate-to-severe CP is intensive. In each session we had 4 active researchers. One researcher ran the computer in charge of data acquisition and

testing parameters, one researcher ran a video camera (to help with interpreting the kinematic data and for future behavior coding projects), and two researchers helped with clinical tests, transfers, adjusting the child in the testing device, communicating with the child and caregiver, and monitoring the child for signs of discomfort or fatigue.

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All subjects sat on a bench while facing forward (Goodworth et al. 2017). For subjects with CP, a movie with audio was playing on a computer monitor located about 85cm in front of the subject at eye level. This helped to entertain children while encouraging subjects to face forward and keep an upright posture. While some small movements could be elicited by the movie, they would not be correlated with the stimulus and would be less than the movements that would occur if children were bored, looking around, or lacking motivation to sit up. Adults listened to an audio story through earphones. All subjects were instructed to maintain an upright posture. An adjustable footrest that tilted with the bench was provided to keep the lower body in a comfortable, neutral, and standardized position.

All subjects were provided a pelvis strap that fixed the pelvis in a vertical alignment to the bench in the sagittal plane(Butler et al. 2010). With only pelvis straps, all segments above the pelvis were free to move. This presented a challenge for most subjects with CP. In 5 subjects with CP, it was possible to safely test trunk posture with the pelvis straps; however, for 2 subjects with CP, it was not safe to complete tests without an additional trunk support located about 5-8cm above the L4/L5 joint (Fig. 1). The trunk support device was described previously (Goodworth et al. 2017). In brief, this trunk support contained horizontal padded arms that moved up and down with the bench, allowing trunk tilt in the frontal plane but preventing translation of the trunk at the point of contact. Each horizontal arm slid up and down on a stationary vertical post with low friction rollers (this vertical post is shown in Fig. 1). Each horizontal arm was also rigidly connected to a second vertical post that was attached to the moving bench (this vertical post was omitted from Fig. 1 for clarity). The horizontal arms moved consistently with the surface up to 5Hz (both magnitude and timing) and the surface to horizontal arm coherence function was essentially one up to 2.5 Hz and 0.97 at 5 Hz (Goodworth et al. 2017). This method allowed the 2 subjects to complete the test while controlling fewer degrees of freedom in the trunk (Butler 1998; Goodworth et al. 2017; Major et al. 2001; Saavedra and Woollacott 2015). The current study focused on postural control at the lowest level where subjects can be safely tested, but we note that kinematic data was also collected at additional levels of support where subjects were more stable and these data will be included in future studies.

209 In this study, we report kinematic results from three trials: quiet sitting (where the bench was stationary), small stimulus (2 degrees peak to peak), and large stimulus (8 degrees peak to 210 peak). These trials were delivered in random order. In the small and large stimulus trials, the 211 bench tilted up and down in the frontal plane according to a pseudorandom ternary sequence 212 waveform, referred to as 'PRTS'. These protocols were previously described in detail 213 214 (Goodworth and Peterka 2009; Goodworth et al. 2017). In summary, the PRTS stimulus drove the surface and bench simultaneously across a range of frequencies. The PRTS stimulus was 215 scaled to one of two different amplitudes (2 and 8 degrees peak to peak). The PRTS waveform 216 217 was 21.78 s long with a fundamental frequency of 0.046 Hz (1 / 21.78 s). For each trial, the PRTS waveform was repeated 11 times in subjects with CP and 5 times in adults. Thus, each 218 219 PRTS trial was about 4 minutes for children with CP and about 2 minutes for adults. By repeating more cycles in subjects with CP, we were able to effectively average out some of the 220 221 variability when calculating average responses to the PRTS. However, both average responses and variability were analyzed and presented in the experimental and modeling results. The analog stimuli were delivered at 200 samples/s.

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Analyses

- 226 Trunk kinematics were measured with a 3D magnetic tracking system (trakSTAR, Northern
- Digital Inc, Waterloo, CA). A sensor was placed at C7 on each subject and the distance to the
- 228 transmitter was approximately 0.6 m. Magnetic tracking was selected for testing this population
- 229 with severe motor impairment because researchers need to be nearby the subject for close
- 230 monitoring and occasional adjustments, which could block markers with a video based
- 231 system(Goodworth et al. 2017). Data collection was 200 samples/s.
- In each subject, trunk tilt with respect to upright in the frontal plane was calculated as the
- inverse tangent of the horizontal translation at C7 divided by the distance between C7 and the
- 234 trunk's axis of rotation. In the 5 subjects with CP who could be tested with just pelvis straps, the
- 235 axis of rotation was L4/L5. In the two subjects with CP requiring external support, the contact
- point of external support with the body was considered the axis of rotation. All adults' axis of
- rotation was considered L4/L5. Trunk tilt with respect to upright was calculated throughout each
- stimulus cycle, and used for all dependent variables. The time based dependent variables were
- 1) average root-mean-square (RMS) trunk tilt across stimulus cycles (referred to as RMS sway),
- 240 2) average RMS velocity across stimulus cycles (referred to as RMS sway velocity, and 3) the
- standard deviation of RMS sway across cycles (referred to as sway variability).
- To obtain a more detailed characterization of the postural control system, we calculated
- frequency response functions (FRFs). The FRF is considered a non-parametric representation
- of the postural system. An FRF decomposes surface tilt and trunk tilt into their frequency
- components and then expresses these in terms of a relative magnitude (gain) and relative
- timing (phase). The detailed calculations of an FRF can be found in previous studies
- 247 (Goodworth and Peterka 2009; Pintelon and Schoukens 2012). FRFs were calculated for each
- stimulus cycle and then averaged over the stimulus cycles. On a logarithmic scale, the higher
- 249 frequency bands contained many more data points than lower frequency bands and these
- 250 higher frequency points had lower signal to noise. Therefore, we averaged more adjacent high
- 251 frequency points together compared to the averaging of adjacent low frequency points, so that
- 252 the final set of FRFs were approximately equally spaced on a logarithmic frequency scale and
- 253 had similar confidence intervals. (Peterka 2002; Goodworth and Peterka 2009; Otnes and
- Enoch 1972). At any frequency, a gain of zero means the trunk is upright and unaffected by the
- surface motion, which is typically considered good for postural control. A gain of one means the
- trunk is moving at the same magnitude as the surface at a particular frequency. A gain greater
- 257 than one means the trunk sway magnitude is exceeding the surface tilt. A phase of zero means
- 258 the trunk and surface are moving in sync at a particular frequency. A phase lag (negative value)
- 259 means the trunk is lagging behind the surface motion and phase lead (positive value) means the
- trunk is moving ahead of the surface at a particular frequency. In response to a sudden
- stimulus, a phase lead would probably be interpreted as the subject anticipating the stimulus by
- 262 moving ahead of it in timing. But during continuous ("steady-state") postural responses,
- 263 modeling studies have shown that phase leads can arise through feedback
- mechanisms(Goodworth and Peterka 2009; Peterka 2003). Our study is focused on identifying
- 265 control mechanisms during continuous steady-state perturbations to balance.

Coherence functions were also calculated. A coherence function indicates the linear relationship between the surface tilt and trunk sway response across frequencies. A coherence of one indicates a perfect linear relationship with no noise (sway variability), no voluntary changes in posture, and no non-linearity's between the surface tilt and body sway(Bendat and Piersol 2011; Peterka 2002). Coherences and FRFs were calculated for each subject and each test session and then presented as averages across sessions in the Results section.

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Model Description

The upper body was modeled as a single link inverted pendulum. This assumption is appropriate to maintain a tractable mechanical system since this is the first modeling study of postural control in CP and multilink models can become extremely complex(Goodworth and Peterka 2018). Using anthropometrics(Winter 2005), the trunk center of mass height and moment of inertia were estimated from Erdman's trunk segment specific regression model(Erdman 1997) after the relative head and arms masses were proportionately scaled based on age(Jensen 1986).

To begin examining control mechanisms that could stabilize the body, we considered that previously identified mechanisms in sitting and standing may also be important for understanding abnormal control in CP. We define control parameters to be all the model parameters other than the inverted pendulum and these parameters could include a Passive and Reflexive mechanism and a Sensory Integration mechanism (van Dieen et al. 2018; Goodworth and Peterka 2009). Our model is shown in Figure 2. The passive component included intrinsic stiffness in the trunk musculoskeletal system that generates torque with no time delay orienting the trunk toward the surface (ie, torque in proportion to trunk tilt deviations from the surface). The reflexive component generates short-time delayed damping orienting the trunk to the bench. Together, the Passive and Reflexive mechanism represents the fast-acting torque contributions from intrinsic stiffness, damping, and reflexive control(Goodworth and Peterka 2009; 2010). The Sensory Integration (SI) mechanism, sometimes termed "active" control, consisted of sensory weights and a neural controller (Bingham et al. 2011; Cenciarini et al. 2010; Goodworth and Peterka 2009; Mergner et al. 2002; Peterka 2002; Welch and Ting 2008). By neural controller, we are referring to the sensory-to-motor scaling: whereas the sensory weights refer to the particular combination of sensory systems the subject is relying on. The sensory weight, Wp, represents sensory cues that orient the trunk to the pelvis like proprioception, and Wv represents vertical orienting cues like vision and vestibular. The sum of Wp and Wv equals 1, so that Wp and Wv represent the sensory reliance and is only one free parameter in the model. The neural controller consists of an active stiffness gain (Ka) that generates torque in proportion to the sensory error (limiting excessive sway), a damping gain (Ba) that generates torque in proportion to the velocity of the sensory error (limiting excessive sway velocity), and an integral gain (Ki) that generates torque in proportion to the sum of sensory error over time (limiting low frequency sway).

The control parameters in the model were determined from an optimization routine ("fmincon") 305 306 (MATLAB, The Mathworks) that selected parameters that minimized the total mean-square-error 307 (MSE) of the normalized difference between the model FRFs and experimental FRFs 308 (Goodworth and Peterka 2009a; Peterka 2002). The fitting routine included lower and upper bounds on parameters which were varied across subjects because of the variability in control 309 mechanisms and because fitting to FRFs could sometimes result in convergence to a local 310 minimum that could be avoided with a minor adjustment to an upper or lower bound (Goodworth 311 312 & Peterka 2018). The typical lower bounds were zero for gains, zero for weights, 10ms for

313 reflexive delay, and 100ms for SI time delay. The upper bounds were 2*mgh for stiffness gains,

314 0.5*mgh for the integral and damping gains, one for sensory weights, 80ms for reflexive time

delay, and 400ms for SI delay.

Depending on the experimental data features, it is possible for some parameters to have

overlapping influences on body sway. For example, if the sensory integration weight (Wp)

- equals one, then the SI stiffness (Ka) and damping (Ba) will orient the body toward the surface
- 319 similar to the Passive and Reflexive mechanism. Therefore, although these two mechanisms
- would still be distinct through their different time delays, they would have some overlap in
- function, which could be problematic with noisy data(Goodworth and Peterka 2018; Pintelon and
- 322 Schoukes 2001). To arrive at a parsimonious model for each subject with CP and for adults, we
- 323 systematically included or excluded model parameters until an accurate and informative set of
- parameters was reached(Goodworth and Peterka 2012), described in detail in the Results
- "Model parameter sensitivity." This approach is consistent with the heterogeneity of CP which
- suggests that each child could be deficient in a different aspect of control (eg, poor damping vs.
- poor sensory integration); and this approach is consistent with the large variability in our
- kinematic data across subjects with CP (Fig. 3). Thus, we did not force each subject with CP to
- have inclusion of the same control parameters. In contrast, there was no reason to believe
- healthy adults would have a significantly different postural control system from each other.
- Further rationale and details of including/excluding parameters is found in the Results sections.
- For completeness, results and interpretation using the full model in CP are included in the
- 333 **Discussion**.
- Time domain simulations with noise were needed to distinguish between the postural response
- and variability. Noise was added to the model (Fig. 2) using MATLAB Simulink. Our noise was
- modeled as a low pass filtered pink noise(Goodworth et al. 2018). In all simulations, the cut-off
- frequency was fixed at 0.01 Hz and determined heuristically (Goodworth et al. 2018; Maurer and
- Peterka 2005). We consider the noise input "sensorimotor" because the variance in torque it
- produces spreads throughout the loops of the model (both sensory and motor). We varied the
- scaling on the noise waveform and report this scaling as a fraction of each subject's body size
- 341 (mass times g times center of mass height, mgh). For reference, a noise scaling of 1
- corresponded to a standard deviation of noise waveform of 0.71 N.m.

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Statistics & model sensitivity

- When possible, summary data was analyzed across subjects. A linear mixed model was used to
- test if stimulus amplitude and test session had a significant effect on RMS sway and RMS sway
- velocity. The Tukey HSD criteria was used for post-hoc pairwise comparisons. A t-test was used
- to determine if model parameters (normalized by body size) were significantly different between subject groups. A p value of 0.05 was the cutoff for significance. Although our sample size is
- subject groups. A p value of 0.05 was the cutoff for significance. Although our sample size small, several important observations were made by calculating a Pearson's correlation
- between a few key variables. We defined R values less than 0.1 as "no" correlation, between
- 352 0.1 0.3 as a "poor" correlation, between 0.3 0.6 as "fair", between 0.6 0.8 as "moderate",
- and above 0.8 as "very strong" (Akoglu 2018).
- 354 Model parameters are generally normalized to each subject's body size (mgh). Correlations of
- model parameters and torque contributions vs. body size are presented. The influence of
- inclusion or exclusion of various parameters is detailed through the MSE and visual inspection
- of model FRFs.

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Results

- 360 Sample time series
- All children with CP tolerated the surface tilt despite being tested with minimal or no direct trunk
- support. Figure 3 illustrates sample data from four children with CP during quiet sitting with no
- surface tilt (left column) and in response to the large surface tilt stimulus (right column). The
- surface stimulus is shown on the bottom row. One apparent finding is the high variability in sway
- during quiet sitting. In some cases, sway during quiet sitting was nearly as high as sway during
- responses to surface tilt stimuli. Despite this variability, it was possible to visually detect the
- trunk tilt response correlating with the stimulus in all subjects. This result was obtained by
- 368 averaging the response across 11 repeated stimulus cycles.
- A wide range in behavior was found across subjects with CP. Two subjects (top two rows) had
- 370 high variability in sway and had large responses to the surface stimuli. In contrast, a third
- subject (third row) had lower variability but a high sway response; and a fourth subject (fourth
- 372 row) showed modest variability with a lower response to the surface tilt stimulus.
- 373 The representative adult subject showed a very small amount of sway compared to subjects
- with CP. This sway was highly correlated with the stimulus, showed minimal variability across
- 375 repeated stimulus cycles, and these sway features were similar across all control subjects.
- 376 Thus, only 5 repeated stimulus cycles were sufficient to obtain reliable estimates of dependent
- variables in adults.

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- RMS sway and RMS sway velocity summary statistics
- 380 Figure 4A shows that stimulus amplitude had a significant effect on RMS sway (P<0.001) and
- RMS sway velocity (P=0.009). RMS sway and RMS sway velocity were 1.15 times higher during
- the small PRTS stimulus vs. quiet sitting; RMS sway and RMS sway velocity were about 1.45
- times larger during the large PRTS vs. small PRTS. Adults had significantly lower RMS sway
- and RMS sway velocity than subjects with CP (p<0.001). In adults, increasing stimulus
- amplitude caused a significant increase in sway responses (p<0.001) and more sensitivity to
- 386 stimulus amplitudes: RMS sway was 4 times higher during the small PRTS vs. quiet sitting and
- 2 times higher during the large PRTS vs. small PRTS.
- We found a fair correlation between RMS sway and age for both quiet sitting and the small
- stimulus trial, and a poor correlation between RMS sway and age for the large stimulus (Figure
- 390 4B).
- 391 RMS sway and RMS sway velocity were not significantly different across repeated sessions
- 392 (Figure 4C). Improved postural control would normally be considered a reduction in RMS sway
- and RMS sway velocity over time. The few subjects who showed monotonic changes in one
- variable either did not show the same change in the other variable or showed minimal overall
- change. The exception to this observation was CP13 whose data is presented over 6 test
- sessions. There was a 50% decrease in RMS sway velocity from the 1st to last session. This
- 397 subject's data is described in more detail in the Discussion.

- and timing (phase). If an FRF is different between subjects, then the postural control system is
- different between subjects. Figure 5 shows the FRFs averaged across sessions for each
- subject. FRFs are ordered roughly from largest sway in which the FRFs were similar between
- 404 the small and large surface tilt stimulus in the top four sets to smaller sway with larger changes
- 405 between surface tilt stimuli in the bottom four sets of FRFs. *Gains*. In the top set, gains at low (<
- 406 0.4) and mid frequencies (0.4-1Hz) were large, typically between 2.5 and 4. This result means
- 407 that the subjects' trunk tilted 2.5 to 4 times more than the surface tilted at these low and mid
- 408 frequencies. At frequencies above 1Hz, gains tended to decrease. There were minimal
- differences in FRF gains between the small and large stimulus amplitudes in the subjects
- 410 presented in the top row.
- The bottom set of FRFs included both the oldest and youngest subjects. At the lowest frequency
- 412 (0.04Hz), gains in the bottom set tended to be smaller (1.2-3) than the top set. As stimulus
- frequency increased, gains generally increased up to about 0.4Hz and then decreased at higher
- 414 frequencies. Increasing surface tilt amplitude (large stimulus) generally decreased FRF gains at
- mid and low frequencies compared to the small stimulus. This result means that these subjects'
- 416 trunk tilted1.2-7 times more than the surface tilted (depending on frequency) when the stimulus
- 417 was small, but were able to limit these trunk tilt responses when the surface tilt amplitude
- 418 increased.
- By comparison, gains in adults were low (0.8-1.2) at 0.04Hz and only slightly increased at
- 420 0.08Hz and then become fairly constant until about 1Hz. At frequencies above 1Hz, FRF gains
- decreased. In adults, there was a clear decrease in FRF gain of about 30-50% in the large vs.
- 422 small surface tilt amplitude.
- 423 Phases. In general, phases were close to zero degrees at low frequencies (< 0.4 Hz) (ie, the
- 424 surface tilt and trunk tilt were moving in-sync) and tended to decrease at higher frequencies.
- 425 meaning that the trunk tilt lagged behind the stimulus at higher frequencies. There were a few
- 426 exceptions where a phase lead or lag was found at the lowest frequency. The phase lag varied
- 427 across subjects from as low as -90deg at 1Hz in one subject to -150deg at 1Hz in another
- subject. There were small differences across stimulus amplitude but no consistent pattern in
- 429 these differences. Adult phase data showed a similar pattern for all adults across frequencies.
- The larger stimulus amplitude resulted in slightly less phase lag between 0.8Hz and 2Hz.
- Coherences. A coherence of one indicated a perfect linear relationship with no noise (sway
- variability) between the surface tilt and body sway. Either a non-linear response or a variable
- response can reduce the coherence. Figure 5 (3rd and 6th row of plots) shows the average
- coherence across sessions for each subject. For the large stimulus, coherences were
- 435 approximately 0.4 to 0.6 at frequencies below 1Hz and decreased at higher frequencies.
- Coherences were lower for the small stimulus (approximately 0.1 to 0.3). There was a lot of
- variance across subjects, with one subject showing coherences between 0.8 and 0.9 for the
- highest stimulus. The two subjects with the highest coherences (CP13 and CP10) correspond
- 439 to the subjects in the 3rd and 4th row in Figure 3. These subjects had relatively low noise (ie, low
- variability in sway). The 2 subjects with the lowest coherences (CP11 and CP23) correspond to
- the 1st and 2nd row in Figure 3, exhibiting high variability in sway. Adults had generally high

coherences between 0.7 and 0.9 at frequencies below 2Hz, with decreases in coherence at frequencies above 2 Hz.

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Model parameter sensitivity

- The FRFs above represent the postural system in non-parametric form. In the following sections, we use a parametric feedback model to investigate the control mechanisms underlying
- 448 each subject's postural system.
- We first included or excluded model parameters until an accurate and informative set of
- 450 parameters was reached. Figure 6 shows the MSE for various model iterations averaged
- between the small and large PRTS stimulus trials. A low MSE represents a good model fit.
- Because some subjects have overall better or worse model fits, we also include a plot of the
- 453 MSE normalized for each subject's "full model" (with all parameters included) in the bottom of
- 454 Figure 6B. This normalized plot lowered the error bars but the overall trend remained the same.
- These trends in subjects with CP (Figure 6B) showed that models with only a Passive and
- Reflexive mechanism (blue) could account for much more of the data than a Sensory Integration
- 457 mechanism alone (red). Because it is conceivable that passive stiffness and passive damping
- 458 (no time delay) could be a viable control system, we also modeled this assumption (green).
- Again, the Passive and Reflexive mechanism (blue) was superior to the model based on
- passive stiffness and passive damping (green). Interestingly, when adding sensory integration
- 461 parameters to the Passive and Reflexive mechanism, there was only modest improvement in
- 462 the MSE.
- However, in looking at the FRFs, there were specific features in each subject's data that were
- 464 not fully accounted for with the Passive and Reflexive mechanism alone. Therefore, all subjects
- with CP required at least one time delayed sensory integration parameter to be added for an
- appropriate fit to the experimental data. Figure 6C left column gain and phase plots provide
- samples of the impact of single mechanism models on the FRF fits in two different
- representative subjects with CP for the large stimulus. Experimental data are black circles.
- 469 Figure 6C right column gain and phases show examples of how adding just one or two time
- delayed sensory integration parameters captures the experimental data better than the model
- 471 based on only a Passive and Reflexive mechanism. These samples also demonstrate examples
- of how adding parameters in the "full model" was typically not necessary to describe the main
- features of the data in each subject with CP.
- 474 Adult data is shown in Figure 6D and 6E. The Sensory Integration mechanism alone was only
- slightly worse than the Passive and Reflexive mechanism alone. But unlike subjects with CP,
- 476 the Passive and Reflexive mechanism did not account for most of the experimental data.
- 477 Incrementally including more parameters improved the model fit to FRFs (Figure 6D). Even
- 478 though the best two parameter Sensory Integration (*Ka+Ba*) plus Passive and Reflexive model
- was only slightly worse than the full model, the benefit of the full model was clear in describing
- 480 the low frequency phase behavior during the small stimulus PRTS (Figure 6E right plot). Thus,
- model fits for adult data included the full model with all parameters included.

Model fits and parameters

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- Model fits for each subject are shown in Figure 7, ordered the same as experimental results in
- Figure 5. Each model was able to account for all the major features of the experimental FRF
- 487 gain and phase plots for both the small and large PRTS surface tilt amplitude, with one
- exception. The model for subject CP24 could not fully capture the high gain values during the
- small surface tilt trial. The pie chart shows the relative torque contribution from each of the
- 490 control parameters averaged between surface tilt trials (described further in the following section
- 491 "Time domain simulations and torque contributions"). Subjects with a more diverse set of control
- 492 parameters contributing to torque generally resembled mature adult data more (eq. CP24 and
- 493 CP10), whereas subjects whose control was dominated by the Passive and Reflexive
- 494 mechanism (CP20 and CP11) were less similar to adults.
- Mean subject model parameters are shown in Figure 8. All stiffness, damping, and integral
- 496 parameters were normalized by each subject's body size (mgh). Subjects with CP had passive
- stiffness (Kp) of about 1.4 times mgh that slightly increased with the larger surface tilt amplitude.
- Reflexive damping (*Bp*) increased from about 0.15 to 0.18 times *mgh* with the larger surface tilt
- amplitude. The sensory weight (Wp) was about 0.85, indicating a high reliance on
- 500 proprioception that did not change between surface tilt amplitude. On average, the active SI
- stiffness (*Ka*) and damping (*Ba*) were about 10 times lower than passive stiffness (*Kp*) and
- reflexive damping (*Bp*). The integral gain (*Ki*) was about 0.1 times *mgh* and slightly increased
- with the larger surface tilt amplitude.
- Nearly all parameters in subjects with CP were significantly different than adults. Subjects with
- 505 CP had much larger contributions from passive stiffness than active SI stiffness; whereas adults
- used active SI stiffness more than passive stiffness. The short latency phasic time delay (τ_0)
- was significantly smaller in CP compared to adults (p=0.018) while the SI time delay (τ_{si}) was
- longer in CP than adults (p=0.001). The sensory weight (Wp), SI stiffness (Ka), SI damping
- 509 (Ba), and reflexive damping (Bp) were all significantly different at p<0.0001. Passive stiffness
- 510 (Kp) and integral gain (Ki) were not significantly different between children with CP and adults.
- In response to increases in surface tilt amplitude, subjects with CP increased their stiffness (Kp)
- and damping (*Bp*) in the Passive and Reflexive mechanism while adults used sensory
- reweighting (Wp decreased and therefore Wv increased) and adults increased their SI stiffness
- 514 (Ka) and damping (Ba) while slightly decreasing their passive stiffness (Kp) and reflexive
- 515 damping (Bp).
- The total stiffness (Kp+Ka) averaged between the two surface tilt amplitudes was slightly larger
- in CP vs. adults but not statically significant (p=0.053); while the total damping (*Bp+Ba*) was
- notably smaller in CP vs. adults (p=0.011) (Fig. 8B). Finally, we explored the correlation
- between total stiffness and damping with subject size (Fig. 8C). The range of sizes in both
- populations was wide with *mgh* ranging from 9.8 to 43 kg.m²/s² in CP and 52 to 96 kg.m²/s² for
- adults. There was a very strong positive correlation between *mgh* and total stiffness for both
- subject groups. In both groups, damping was also positively correlated with mgh, but less so
- 523 than stiffness.
- 524 Correlations between age and model parameters in subjects with CP were typically poor or fair:
- 525 Ka (R=0.32), Ba (R=-0.26), Ki (R=-0.65), SI time delay (R=-0.06), Kp (R=-0.42), Bp (R=-0.47),
- reflexive time delay (R=0.53). Wp was not included because only 1 subject with CP had a non-

unity value. The two highest correlations (*Ki* and reflexive time delay) indicates that older subjects with CP tended to have less integral gain control and had longer reflexive time delays.

Time domain simulations & torque contributions

Figure 9 shows sample time domain simulations of two subjects with CP (one with low noise and one with high noise) and one representative adult. Simulations in the time domain provide an intuitive way to see quality of model fit at low frequencies because stimulus and response amplitudes are largest at the lowest frequencies. These simulations also provide a means to quantify the various torque contributions to postural sway. Figure 10A shows the various torque contributions from each control parameter for one representative adult and one subject with CP. In all subjects, torque from the stiffness parameters provides torque in the opposite direction of the surface tilt and trunk tilt to resist extreme deviations from upright. Damping control parameters generate higher frequency torque and this torque changes quickly. The integral control parameter generates torque at a very low frequency and this torque changes slowly in the opposite direction of trunk tilt and surface tilt. The sample subject with CP had a model that could describe his data without the need for *Ka* and *Ba*. To obtain a summary metric, we calculated the standard deviation of each torque contribution. Then, we quantified the relative contribution of these torque metrics from each control parameter and present this next to each subject's model FRFs in Figure 7.

Figure 10B shows the sum of each standard deviation torque contribution normalized to *mgh*. Subjects with CP had over 2 times larger torque contributions during the large stimulus. A scatter plot of all subjects showed a very strong correlation between RMS sway and the sum of torque contributions. These results are expected because greater deviations from upright result in larger torque due to gravity and therefore greater deviations from upright require greater corrective torque.

Inclusion of noise in simulations

To explore the high variability in sway across stimulus cycles in subjects with CP, we inserted sensorimotor noise into the time domain simulation. For each subject, we varied the noise amplitude until the RMS sway and sway variability was within 20% of the experimental data. In the results shown in Figure 11A, the difference between model and experimental data was 9% (averaged across subjects), with a maximum of 13.5% in one subject. Data in Figure 11 are ordered from least to greatest scaling on noise, meaning that subjects farther down in the plot required the highest level of noise to be included in their postural system to accurately capture the variability found in their experimental data. Noise levels in each subject were only identified during the large surface tilt condition because the large PRTS condition had higher signal to noise than the small surface tilt (eg, coherences in Figure 5) and we were therefore more confident in the control parameters identified during the large stimulus trial. In addition, there were very strong correlations in sway variability between large and small surface tilt trials (R=0.93) and between large surface tilt and quiet sitting (R=0.83), Figure 11B, suggesting a common noise source within each subject.

Several results emerge from this analysis. First, it was possible to accurately model each subject's postural response and variability through a linear parametric feedback model with the addition of non-linear sensorimotor noise. Note the similarity between average cycle trunk tilt and 95% confidence intervals and the similarity in the four sample subjects showing the response to 11 cycles to those in the experimental data in Figure 3). Second, noise levels varied greatly across subjects, with noise scales ranging from 0.01 to 0.19 across subjects. Third, the inclusion of noise further increased the torque required to stabilize the body. With the inclusion of noise, the average corrective torque in subjects with CP increased to levels 2 to 3 times higher than those reported in Figure 10B. Fourth, there was only a fair correlation between noise level and RMS sway (R=0.35) during the high stimulus condition (Fig. 11C, left). This means that a large average response to the stimulus was likely due to control parameters (eg, stiffness and damping). In contrast, the correlation between noise level and RMS sway increased to moderate for the small stimulus (R=0.6) and quiet sitting (R=0.71), Fig. 11C, middle. This result means that subjects with higher noise levels swayed more in quiet sitting. This result may be expected since sensorimotor noise is often considered the primary component that triggers spontaneous sway in the absence of an external stimulus. Finally, we found a moderate negative correlation between noise and age (R=-0.75, Fig. 11C, right) meaning that older subjects tended to have less sensorimotor noise in our study, consistent with the negative correlation coefficient found between spontaneous sway and age in Figure 4B.

Discussion

This current study outlines a specific way to view trunk postural control in severe CP. We hypothesize that the postural control system in severe CP is largely based on a Passive and Reflexive mechanism along with sensorimotor noise. Control parameters contributed to abnormally large average responses to perturbations and the interaction of sensorimotor noise with control parameters contributed to large sway variability across repeated stimuli. This response and variability distinction has meaning for treatment because it points to two different causes for large postural sway. We found control parameters, sensorimotor noise, and sway patterns varied widely across our 7 subjects with CP age 2.5 to 13 years. Positive correlations between sensorimotor noise level and postural sway (RMS) were highest during quiet sitting and lowest during the large surface tilt stimulus. Age had a strong negative correlation with sensorimotor noise.

In general, the Passive stiffness and Reflexive (damping mechanism could account for much of the response to perturbations for children with CP. In 5 out of 7 subjects with CP, changes in control between stimulus amplitude were small, suggesting deficits with adaptive postural control. Moreover, the changes between stimulus amplitude that did occur were primarily accounted for through an increase in stiffness and damping in the Passive and Reflexive mechanism, demonstrating a high reliance on intrinsic stiffness and proprioceptive sensory input. In contrast, adults showed a high use of sensory integration and reliance of vision and vestibular feedback that oriented the trunk more vertical. Adults also demonstrated sensory reweighting toward vision and vestibular feedback and away from proprioception at larger surface tilt amplitudes. With larger surface tilt amplitudes, adults also used more stiffness and damping from the Sensory Integration mechanism and less stiffness and damping from the Passive and Reflexive damping mechanism (Figure 8A, top row left). The modeling results indicated that our test population with CP had higher overall stiffness and significantly lower

damping than the adult postural control system. In subjects with CP, the surface-orienting stiffness and damping required subjects to generate much higher corrective torque than adults and these torque contributions were further amplified by the child's sensorimotor motor noise.

Clinical implications

Historically, postural control studies and outcome measures have focused on ambulatory children with CP. They required the ability to sit or stand independently which excluded children with deficits in trunk postural control from participating. There are few effective interventions for postural control in CP and the studies that have demonstrated evidence of effectiveness were conducted primarily with ambulatory children (Dewar et al. 2015; Fowler et al. 2007; Novak et al. 2013; Saavedra and Goodworth 2019). It is difficult to imagine how to begin addressing this clinical gap when almost nothing is known about the underlying postural control system in nonambulatory children with CP. The ideas and modeling hypothesis put forth in this study are therefore an important starting point to understanding the presence and absence of postural control in severe CP.

Simpler control in moderate-to-severe CP. Subjects with CP typically had a simpler control system with significant torque contributions from the fast-acting Passive and Reflexive mechanism. Torque from this mechanism comes from inherent trunk stiffness and damping, neural-modulated tone, and reflexes. This type of control could be considered more stable because it does not involve the long time delays associated with the Sensory Integration mechanism (Antritter et al. 2014; O'Brien et al. 2015). This fast-acting control is also considered more simple and foundational. It has been suggested that infants use passive control processes prior to developing the more refined adaptive control(Hadders-Algra et al. 1998). It has also been shown that sitting trunk responses to a perturbation were less consistent in children with more severe CP compared to less severe and latency for response onset was longer for children with CP compared to typically developing peers (Brogren et al. 2001). Consistent with these ideas, we found the subject with the most similar sensory integration to adults (CP10) scored best on the clinical GMFCS (level III), suggesting the most developed control system.

We also found a range in torque contributions amongst the subjects with CP (Fig. 7) suggesting a range in complexity of motor control across our subjects. But nearly all subjects with CP could be considered to have less complexity than mature control based on the variety of torque contributions present (Fig. 7). Interestingly, researchers have investigated muscle synergies in ambulatory individuals and found reduced complexity is associated with CP (Steele et al. 2015) and adults following brain damage from a stroke(Clark et al. 2010). More complexity of muscle synergies has also been linked to better surgical outcomes in CP(Shuman et al. 2019; Steele et al. 2015). One downside of fast-acting Passive and Reflexive control is that this mechanism tends to orient the body perpendicular to the bench, and thus away from upright when the bench tilts. That is, when the bench tilts, muscles and tendons on the downward side of the body are stretched. These stretched muscles and tendons generate torque through passive and reflexive forces that resist the stretch by moving the body perpendicular to the tilted surface. Once oriented away from upright, gravity generates a torque that further accelerates the body away from upright(Peterka 2002). We found a high tendency for children with CP to orient toward the surface with the fast-acting Passive and Reflexive mechanism and in 6 out of 7 subjects with CP the Sensory Integration mechanism included a very high reliance on proprioception which also

orients to the surface (Wp = 1). In these 6 subjects with Wp=1, we found moderate and fair negative correlations between RMS sway and total stiffness, damping, and integral gain (Figure 12A, white circles). This means that, without using vision or vestibular feedback, subjects with the highest RMS sway tended to show low corrective control (low stiffness, damping, and integral gains). Only through increasing stiffness, damping, and integral gains were subjects with CP able to limit postural sway when Wp=1. Not surprisingly, increasing stimulus amplitude was compensated for with an increase in total stiffness, reflexive damping, and integral gains (Figure 8A). Figure 12 also illustrates that the one subject with CP (CP10) who relied upon vision and vestibular cues exhibited the lowest RMS sway with the lowest stiffness, damping, and integral gains, and consequently the least torque generation relative to body size.

Together, the large deviations from upright in children with CP required much higher corrective torques than adults. Our models with sensorimotor noise suggested that children with CP generated 3 to 5 times more torque than adults (normalized to their body size). This finding is in agreement with clinical observations that children with moderate-to-severe CP are often fatigued during the seemingly "simple" act of maintaining a vertical posture. Thus, not only do visual and vestibular cues help limit sway relative to the base of support and provide a stable visual field, these cues also help reduce the overall torque (and presumably energy) needed to maintain balance by keeping oneself mostly upright. However, ability to rely on these sensory systems requires a Sensory Integration mechanism that can interpret these sensory cues and appropriately scale the sensory-to-motor outputs in the presence of time delays, which is often impaired with neurological damage(Horak and Diener 1994; Kim et al. 2009). For many non-ambulatory children, most of their time is spent in a supported and semi-reclined wheelchair that offers little need or opportunity to practice using these sensory cues for postural control.

Response vs. variability. An observation of quiet sitting is the simplest way to assess trunk control in the clinic. But in quiet sitting, response and variability are intertwined. In quiet sitting, sensorimotor noise is the stimulus that the control system responds to, making it very difficult to know if large postural sway comes from high sensorimotor noise or a poorly calibrated control system. In viewing postural sway, we distinguish response from variability. As others have emphasized(Pasma et al. 2014; van der Kooij et al. 2005), it is necessary to use an external stimulus to tease apart these two important aspects of the trunk postural system. The sway response comes directly from the postural control system (eg, stiffness and damping) and this dictates the average response to a stimulus. When an external stimulus is used (such as PRTS in our study), then an estimate of the response can be made through adequate averaging. In contrast, variability comes from the uncertainty in both sensory cues and processing and variability in muscle activations, together termed sensorimotor noise (Faisal et al. 2008; Goodworth et al. 2018; van der Kooij and Peterka 2011). Sensorimotor noise causes the variable response to an external stimulus and can be separated from the average response. The SATCo is an example of a clinical test that attempts to quantify quiet sitting ability separately from reactive control (response to a manual push perturbation)(Butler et al. 2010). In standing, the BESTest is an example of a clinical test that attempts to distinguish these skills(Horak et al. 2009).

This distinction between response and variability has meaning for treatment as well because it points to two different causes for large postural sway. We found subjects with large sensorimotor noise were not necessarily the same subjects with a large response to stimuli. Knowledge about this population is so limited, it is premature to suggest specific interventions.

However, we note interventions focusing on reducing high sensorimotor noise would certainly differ from interventions that focus on rescaling control parameters or shifting sensory reliance to vertical orienting sensory cues. Physical rehabilitation in severe CP is very labor and time intensive, thus, it is imperative to maximize efforts with the most targeted and subject specific approaches as possible.

Finally, we point out one result that illustrates the complexities of closed loop control. We found subjects with larger RMS sway (mean response across cycles) also had large sway variability across repeated stimulus cycles for both the high and low PRTS stimulus (Figure 12B). At first, this result was counter-intuitive because Figure 11C shows that RMS sway was not notably correlated with sensorimotor noise (the source of variability across repeated cycles). The answer was only evident when considering the control system parameters of the 6/7 subjects with CP who did not use visual or vestibular feedback (*Wp*=1). In these subjects, the highest RMS sway was associated with the lowest corrective control (low stiffness, damping, and integral gains), Figure 12A. From a modeling viewpoint, low corrective control parameters allow sensorimotor noise to have a relatively larger impact. Therefore, a subject with high RMS sway could have a relatively low sensorimotor noise level that causes high variability due to an "under responsive" control system. Taken together, sensorimotor noise creates variability in postural responses but the magnitude of these variable responses are a function of both sensorimotor noise levels and the control system parameters that respond to it.

Long term motor learning

We further analyzed the subject who showed the most consistent changes in RMS metrics across repeated sessions (CP13 in Figure 4C). This subject showed monotonic decreases in RMS velocity across sessions. We averaged the first three sessions and the last three sessions and present each FRFs and model fit in Fig. 13. The FRFs differed mostly in the low-to-mid frequencies where the first 3 sessions showed larger gains. The parameters indicated this subject did not adjust total stiffness relative to his body mass but instead increased his total damping across sessions. High gains across a range of frequencies can be caused by insufficient damping(Goodworth et al. 2018; O'Brien et al. 2015). In fact, across all subjects with CP, we found significantly lower overall damping (Fig. 8) which may be related to a published finding that ambulatory children with CP take 2-3 times longer to stabilize their center of pressure following a perturbation (Burtner et al. 2007). In subject CP13, the torque distribution indicated the increased damping across time came from the Sensory Integration mechanism, Ba. In addition to the increased active damping, there was a slight decrease in Ki that mostly affected the low frequency body sway. Other model parameters were similar across sessions and his sensory reliance remained on proprioception. Together these results suggest some modest improvement was possible in this subject who was able to learn to use damping control of sensory feedback to a greater extent with age. This subject was tested over a span of about 2.5 years. Growth was observable as a major change for him during this period of time. Increased damping could potentially result from soft tissue deformation related to waist circumference. We did not take waist measurements for this child. But based on changes in this subject's height and weight, we are able to calculate his BMI which changed from 15.5 to 17.6 across the 2.5 years of this study. Thus, he remained underweight and was unlikely to have adequate change in waist circumference to account for his improved damping. He was also engaged in a Targeted Training home program across these sessions(Butler 1998; Curtis et al.

2016), where he showed improved SATCo for static control and reactive control. The improved reactive control during the clinical test (ie, appropriate response to a sudden trunk push) is consistent with his increased damping to better respond to the PRTS stimulus.

Comparison to other studies

Age-related effects. Several previous studies have shown postural sway is higher in younger children and decreases toward adult levels around the age of 12 years old(Bair et al. 2007; Cuisinier et al. 2011; de Sá et al. 2018; Peterson et al. 2006; Polastri and Barela 2013). In the current study, we believe age can account for some of the differences in postural sway between children with CP and adults; but we suggest most of the differences can be attributed to CP itself. Most analogous age-related postural studies have been performed in standing, so comparisons are difficult, but some observations are made below. We also discuss the challenges of a true control population for severe CP in the *Limitations* section.

Previous studies comparing standing postural sway at different ages in typically developing children and healthy adults have shown notable correlations between age and standing postural sway(Cuisinier et al. 2011; de Sá et al. 2018; Peterson et al. 2006; Polastri and Barela 2013). In contrast, we found only poor or fair correlations between age and RMS sway in the current study (Fig. 4B). In previous studies, frontal plane center of pressure movements were about 1.4-1.5 times higher in 5 and 7 year-olds compared to adults during quiet standing(Cuisinier et al. 2011; de Sá et al. 2018). In the current study, these differences were much larger, about 20 times and 9 times higher for the average subject with CP's RMS sway and RMS sway velocity vs. adults, respectively (Fig. 4A). When a visual stimulus was provided in a previous study, 4 year-olds swayed about 2 times more than adults(Peterson et al. 2006). In the current study, these differences were again larger. The average subject with CP responded to the large bench tilt stimulus with 3.7 and 3.9 times larger RMS sway and RMS sway velocity, respectively, compared to adults. Previous studies have provided evidence that children 4 years and older can use sensory reweighting during postural tasks (Bair et al. 2007; Polastri and Barela 2013) but that the magnitude (or ability) is not as strong in younger children compared to adults(Bair et al. 2007; Cuisinier et al. 2011; de Sá et al. 2018; Peterson et al. 2006; Polastri and Barela 2013). In the current study, we found very little evidence for any sensory reweighting between support tilt stimulus amplitudes in subjects with CP (Wp was constant in Fig 8A upper right) whereas adults demonstrated sensory reweighting.

Despite these differences, we found some experimental evidence in our study that age played a role in the elevated sway. We found sensorimotor noise was moderately correlated with age (Fig. 11C, right), suggesting that younger children had more variability in their control system in our study. These differences may have been most important during quiet sitting where R values relating age to RMS sway (Fig. 4B, left) and age to sensorimotor noise (Fig. 11B, middle) tended to be higher than when an external stimulus was provided. The larger variability in quiet sitting in younger children could be influenced by normal development (as younger children have less experience with postural control) or influenced by a shorter attention span and less ability to focus on the balancing task. Variability has been shown to be a key parameter of typical development and has been related to explorative activity during which the sensorimotor responses are refined and shaped with adaptation to changing environmental and task constraints(Adolph et al. 2015; Hadders-Algra 2010). It can also be related to development of

attentional focus. The younger children in this study were more likely to periodically switch their focus from the video to their parent or the researcher while the older children tended to remain visually focused on the video.

Analyzing variable movements in CP. One unique aspect of these data was the high variability compared to those previously published in adult populations using similar techniques. To account for this variability, we typically averaged across at least 30 stimulus cycles (11 per session times 3 sessions) for subjects with CP; whereas adults only required about 5 repeated cycles of the high stimulus amplitude. Using a high stimulus amplitude and averaging across many cycles prior to modeling is consistent with findings from a previous methodological study looking at system identification in posture and the influences of noisy data(Goodworth and Peterka 2018).

Other published models. The model in the current study consisted of the same Passive and Reflexive mechanism and Sensory Integration mechanism published previously for the trunk postural control system(Goodworth and Peterka 2009b; 2010). Model parameters in adults (comparable population) were similar to those reported previously(Goodworth and Peterka 2009). Very similar or identical mechanisms have been published in other studies of postural control(Cenciarini and Peterka 2006; Mergner et al. 2002; Peterka 2002). The Goodworth and Peterka models(Goodworth and Peterka 2009; 2010) included a medium latency reflexive mechanism that was not included in the current model. The current model did not have as high of a stimulus frequency bandwidth and this medium latency mechanism could not be consistently identified using the narrower bandwidth in the current study. The model in the current study included a simplified representation of sensorimotor noise as a single input. While several modeling studies have used a similar method(Boonstra et al. 2013; Maurer and Peterka 2005), it is possible to add detail by considering more than one source(Goodworth and Peterka 2018; Goodworth et al. 2018; van der Kooij and Peterka 2011).

Also, a few existing posture models have included additional detail by representing muscle activation dynamics (eg, Kiemel et al. 2007; van Dieen 2018). Muscle activation dynamics refers to the mV to N.m transformation in muscles that act across a joint. The current model approximated sensory to torque scaling as a single reflex and neural controller process. As such, muscle activation dynamics were not separately represented, but rather lumped within each of these processes. These lumped time delays are inclusive of the muscle activation dynamics. If activation dynamics were included in the current study as a low pass filter (Kiemel et al. 2007; van Dieen 2018), the time delays would only represent the sensory to mV process and would be shorter than those currently presented. However, unless muscle activation dynamics can be estimated a priori, inclusion of this transformation would introduce additional model parameters that may not have been reliably estimated with our variable kinematic data. At present, very little is known about trunk muscle activation dynamics in severe CP. However, several studies have found heightened co-contraction in CP which would impact muscle activation dynamics and passive stiffness. Specifically, trunk co-contraction (simultaneous activation of trunk extensors and rectus abdominus) was found during sitting perturbations (Brogren et al. 1996, van der Heide and Hadders-Algra 2005) and in ambulatory children with CP during standing perturbations (Nashner et al 1983, Burtner et al 1998). High levels of cocontraction would be consistent with modeling results in the current study showing large passive stiffness in children with CP (Fig. 8A, 2nd row).

Finally, other researchers investigated trunk postural control in adults through experimentation and modeling and found stronger evidence for control based on intrinsic and reflexive mechanisms that also included acceleration feedback, and they found weaker evidence for sensory integration (especially vestibular) (Maaswinkel et al. 2015; van Dieen et al. 2018; van Drunen et al. 2016). Experimental design (different perturbation direction, stimulus bandwidth, and stimulus modality) probably accounts for most of these differences to our study. But the fact that adults can incorporate a variety of mechanisms depending on the circumstance is noteworthy as it points to the flexibility available in healthy control. Thus, with a different experimental design that promotes primarily intrinsic and reflexive control, we may find more similarities in control processes between subjects with CP and healthy controls. Future modeling could also investigate acceleration feedback.

The Full Model in subjects with CP

Our modeling is a first testable hypothesis about how abnormal postural control takes place in CP. As such, we cannot completely rule out alternative hypotheses. For example, we analyzed the parameter values obtained from model fits using the "full model" of CP (inclusion of all parameters) to those presented in the Results section that included few parameters. The proprioceptive weight, integral gain, and time delay values were similar between the full model and the model presented in the Results section. The SI stiffness and damping converged to larger values in the full model but still lower than adults. The most notable difference was in the reflexive damping control parameter, which was almost 2 times higher in the full model compared to the model presented in the Results section. A sensitivity analysis suggested that this larger fast-acting damping helped to stabilize the system when more time delayed SI torque was generated in the full model.

In our view, there are several reasons the full model was reasonable for adults but was inferior for subjects with CP compared to the models presented in the Results section that included fewer parameters: 1) Six out of 7 subjects with CP had a proprioceptive weight of one. Thus, the only difference between stiffness and damping in the SI mechanism (orienting to the surface) vs. Passive and Reflexive mechanism (also orienting to the surface) is the time delays. Because of this redundancy, in the full model, many subjects with CP had inconsistent converging of parameters where stiffness in one mechanism could be interchanged with stiffness in another mechanism with little impact on the results. In contrast, adults typically sway less than the surface and therefore the SI mechanism included a very high visual and vestibular weight (orienting toward vertical) that was more distinct from the Passive and Reflexive mechanism (orienting to the surface and bench). 2) In the process of examining the single mechanism model, the MSE in the Passive and Reflexive mechanism in CP was always far better than the Sensory Integration mechanism. Also, the Passive and Reflexive mechanism alone was not much different than the full model. Generally, fewer model parameters that describe the data equally well is preferred because it increases confidence in parameter estimation and reduces the chance for redundancy in parameter function(Goodworth and Peterka 2018; Pintelon and Schoukens 2012). In contrast, the adult data showed almost equal MSE with the Sensory Integration mechanism alone vs. Passive and Reflexive mechanism alone; and the MSE notably decreased with inclusion of both these mechanisms. 3) The CP models presented in the Results are consistent with studies suggesting that passive and reflexive control develops first, whereas sensory integration and sensory reweighting is more advanced and may be a more mature

postural control mechanism(Bair et al. 2007; Hadders-Algra et al. 1998; HaddersAlgra et al. 1996; Shumwaycook and Woollacott 1985)

Limitations & future research

Our adult population was a convenient sample to test and provided an important and clear benchmark for mature trunk postural control. We normalized all parameters to body size. However, one clear limitation is the lack of a true control population for these children with CP. Finding a true control population is difficult to conceive. For example, age-matched controls have had very different experiences and practice using their postural muscles, sensory cues, and interacting with gravity. In addition, since our population of subjects with CP ranged from 2.5 to 13, age-matched controls would need to be a larger sample than the present study to provide averaging of multiple control subjects for each age range (Polaski & Barela 2013, de Sá et al. 2018; Cuisinier et al. 2011; Peterson et al. 2006). In our view, typically developing infants would provide another informative comparison. Typically developing infants learn to control their entire trunk by gradually gaining postural control over the head first and then the upper trunk and then lower trunk which generally matches the level of severity in CP(Saavedra et al. 2012; Saavedra and Woollacott 2015). Infants could be matched to subjects with CP by their level of trunk control (Table 1, right column) which would make for a more similar experience with gravity and more similar function level to more directly isolate the effects of neural damage with CP during development. However, infants have different cognitive development and less experience dealing with postural deficits.

A second limitation is that we only investigated the lowest level of support where subjects could be safely tested. This task was sufficiently complex for a single study and this was the most intuitive approach for a baseline understanding of impaired sitting. In the future, postural data with different levels of trunk support can be analyzed and we expect subjects to show better trunk control consistent with their SATCo(Saavedra and Woollacott 2015). Third, it was not possible to consistently test subjects with CP with eyes closed because most of these subjects would not tolerate this test condition. Comparing eyes open to eyes closed (or providing a visual stimulus) would be a good way to test our hypothesis that 6 out of 7 subjects with CP did not make use of visual feedback for postural control (Bair et al. 2007). Providing different stimulus amplitudes in eyes closed is a good way to test sensory reweighting and our hypothesis that most subjects with CP did not make use of vestibular feedback(Peterka 2002; Goodworth & Peterka 2009).

Conclusion

What postural control mechanisms are used by non-ambulatory children with CP? In our set of 7 subjects, our study points to two things: 1) most children with CP had a simpler Passive and Reflexive control system with only modest contributions from a Sensory Integration mechanism (whereas adults had a major contribution from Sensory Integration), 2) The Sensory Integration mechanism for children with CP included only proprioceptive feedback and not visual or vestibular feedback for 6 out 7 subjects (all GMFCS levels IV and V), whereas adults were able to flexibly reweight their use of proprioceptive, visual and vestibular feedback 3) children with CP showed much higher sensorimotor noise. This noise had a moderate negative correlation

with age. Together, these features of postural control created abnormally large postural sway which in turn resulted in abnormally large generation of corrective torque. Since these children have limited opportunities to experience and practice vertical upright control, it is unknown if these deficits are a direct result of neural damage or if they are a secondary, developmental limitation, from disruption of activity-dependent processes for learning and integration of postural control.

Figure Legends

- Figure 1. Experimental design to study typical and atypical trunk postural control. A) A surface tilt stimulus was used to evoke trunk tilt postural responses in sitting subjects. Surface tilt and bench tilt were synchronous. B) Photos of three subjects. The pelvis was stabilized in the sagittal plane with pelvic strapping. When needed, trunk support just above the pelvis was provided, which moved up and down with the bench but prevented horizontal translation of the trunk at the point of contact.
- Figure 2. Closed loop feedback model of the trunk postural control system. *J* is the trunk's moment of inertial above L4/L5 or the level of support; *mgh* is the trunk's mass x g x center of height above L4/L5 or the level of trunk support; *s* is the Laplace variable; control parameters are represented as K (stiffness), B (damping), and Ki (integral); e^{-r*s} is a time delay; *Wp* and *Wv* represent the proprioceptive and vestibular+visual weights, respectively. In certain modeling analyses, sensorimotor noise (low pass filtered pink noise) was added to investigate sway variability across repeated stimuli. See text for more modeling details.
 - **Figure 3.** Sample time series showing four subjects with moderate-to-severe cerebral palsy (CP) (1st-4th row), one sample adult (5th row) and the repeated surface tilt pseudorandom stimulus (PRTS) (bottom row). The adults only required 5 repeated stimulus cycles to confidently average responses since the adults were much less variable than subjects with CP. The top two subjects (CP23 and CP11) show highly variable movements during quiet sitting (no stimulus) and show large and variable responses to the PRTS stimulus. CP13 (3rd row) showed low variability but high responses to PRTS while CP10 (4th row) had the lowest responses and variability of all subjects with CP.
 - **Figure 4.** A) Zero-meaned root-mean-square (RMS) sway and RMS sway velocity of the trunk tilt averaged across subjects for different stimulus conditions. For each subject, the RMS sway was first calculated for each stimulus cycle and then averaged across cycles. Error bars represent SD across subjects. B) Correlations between RMS sway and age for the different stimulus conditions. C) RMS sway and RMS sway velocity across test sessions. Most subjects had data for two or three sessions while one subject (CP13) had data for six sessions.
 - **Figure 5.** Mean FRFs for each subject with cerebral palsy (CP) and the mean adult FRF (bottom right). A small gain indicates a small deviation from upright relative to the surface tilt and is typically considered "good" postural control. For reference, the dotted line is placed at a gain of 2 and phase of 0. Subjects with CP were ordered from upper left to bottom right as those least resembling adults (exhibiting high gains and little change in FRFs between stimulus amplitudes) to those more resembling adults. In adults, error bars on gain and phase are the

- average 95% confidence interval for each subject and the error bars on coherence are 1SE across subjects.
- Figure 6. A) Simplified schematic of the two control mechanisms of the model to aid in the
- interpretation of the plots in this figure (the entire feedback model is shown in Figure 2). B)
- 970 Model accuracy in subjects with cerebral palsy as a function of inclusion or exclusion of specific
- 971 mechanisms and parameters. Accuracy was quantified with the mean square error (MSE)
- between model and experimental frequency domain data, where a low MSE indicated a better
- 973 fit. MSEs are averaged between the small and large stimuli. Since some subjects had better or
- worse fits across all iterations, the 2nd MSE plot normalizes to each subject's full model MSE. C)
- 975 Sample data showing how FRFs are influenced by inclusion or exclusion of different
- 976 mechanisms and parameters. Two subjects with CP are shown for the large PRTS stimulus. D)
- and E) MSE and sample FRFs for the mean adult data.
- 978 Figure 7. Model FRFs (solid lines) and experimental FRFs (symbols) for each subject with
- 979 cerebral palsy (CP) and the average adult (bottom right plot). The ordering of subjects with CP
- 980 matches those of Figure 5 and generally follows least (upper left) to most similar to adult. In
- 981 CP20 & CP11, it was only possible to fit the model to 1.6Hz (as opposed to 4Hz in the other
- 982 subjects). The pie chart represents the relative torque contribution from the active Sensory
- Integration (SI) mechanism (Ka, Ba, and Ki) and the Passive and Reflexive mechanism (Kp and
- 984 Bp).
- 985 Figure 8. A) Mean model parameters for subjects with cerebral palsy (white circles) and adults
- 986 (black circles) as a function of surface tilt (stimulus) amplitude. Each subject's parameters were
- normalized by their body size (*mgh* = mass times g times center of mass height of the trunk
- segment). Error bars are 1SE across subjects. Note, that Wv = 1 Wp. The total stiffness is Kp
- + Ka and total damping is Bp + Ba. B) Total stiffness and damping as a function of body size.
- 990 Figure 9. Model parameters identified from fits in frequency domain were used to simulate the
- 991 trunk tilt response to the surface tilt in the time domain using MATLAB Simulink. Model
- 992 predicted responses (solid black line) and the mean experimental sway (grey line) for one adult
- 993 subject and two subjects with CP one with low variability (small 95% confidence interval
- 994 across repeated stimuli) and one with high variability.
- 995 **Figure 10.** A) Time domain simulations of the torque generated from different control
- 996 parameters illustrate how stiffness, damping, and integral control contribute to trunk tilt in two
- 997 representative subjects. B) The total torque (standard deviation) from all parameters averaged
- 998 across subjects (1SD) and normalized to body size (mass times g times center of mass height,
- 999 mgh) for the large stimulus condition (left). A scatter plot of total torque vs. root mean square
- 1000 (RMS) sway of the trunk tilt demonstrates that larger trunk deviations from upright require more
- 1001 total corrective torque to resist falling over.
- Figure 11. Simulated time series with sensorimotor noise for subjects with cerebral palsy (CP).
- 1003 A) Simulated time series for each subject with CP (left) to the large stimulus (bottom waveform).
- Subjects are ordered from least to most sensorimotor noise required in the model. Experimental
- time series for several of these subjects was shown in Figure 3. The average cycle response
- 1006 with 95% confidence intervals across the repeated 11 cycles for both model and experiment
- 1007 (right). B) Sway variability during the high PRTS stimulus is highly correlated with sway
- variability during guiet sitting and the low PTRS stimulus. C) Relation between sensorimotor
- noise (horizontal axis) and RMS sway during large stimulus, no stimulus, and age (vertical axis).

Figure 12. A) Control parameter values vs. RMS sway in subjects with CP illustrates that higher 1010 stiffness, damping, and integral gains are required to limit postural sway and orient the body 1011 toward the tilting surface when Wp=1 (white circles). The one subject with CP with high reliance 1012 on vision+vestibular was able to limit sway with lower stiffness, damping, and integral gains. B) 1013 RMS sway vs. sway variability for PRTS stimulus conditions in subjects with CP. 1014 1015 Figure 13. Motor learning in the one subject with CP (CP13) who demonstrated monotonic decreases in RMS sway velocity across time. The second half of sessions (blue) showed 1016 smaller gains at mid-frequencies due to higher damping. Model fits are solid lines and 1017 1018 experimental data are symbols. 1019 1020

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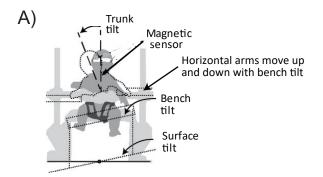
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Table 1. Anthropometric and clinical test summary for subjects with cerebral palsy.

subject	Age (months)	Height (cm)	Weight (kg)	GMFCS level	SATCo
CP10	128-140	136-139	36-42	III	LT to UL
CP 11	52-64	99-110	14-16	IV/V	MT
CP 13	128-160	122-135	23-32	IV	UT to MT
CP 15	52-57	109-110	16-16.2	V	MT
CP 20	55-70	99-99	14-17	V	MT
CP 23	25-43	88-104	12-18	IV	UT to LT
CP 24	24-39	99-98	15-16	IV	MT to LT

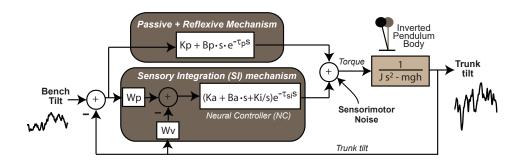
Note, each subject with CP was tested across multiple sessions separated by 2 - 6 months. Anthropometric and clinical test ranges are shown from the first to last session. GMFCS was measured and confirmed during each session. The SATCo lists the most inferior level of external support where subjects demonstrated static, active, and reactive control. Four subjects changed their SATCo scores from the first to last test session. UT = upper-thoracic; MT = mid-thoracic; LT = lower-thoracic; UL = upper lumbar

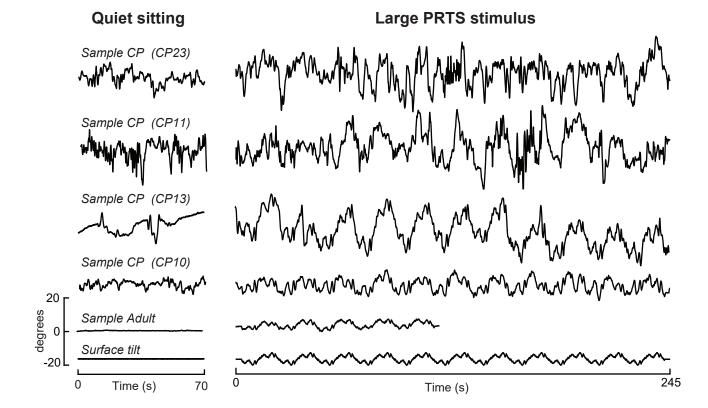


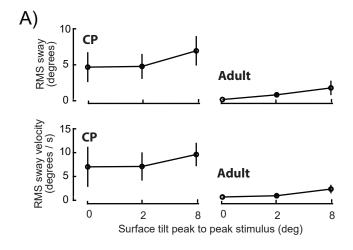


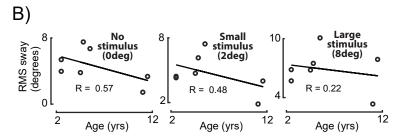


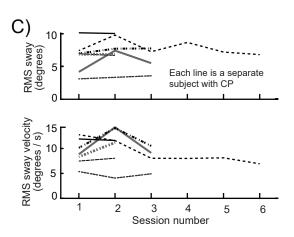


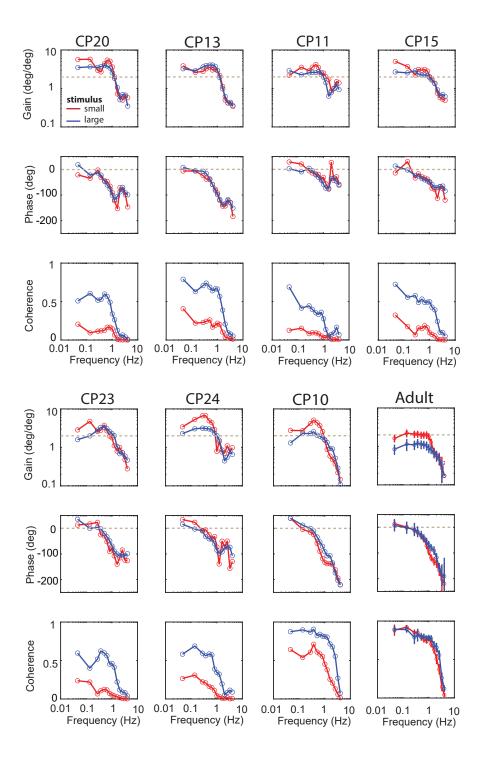




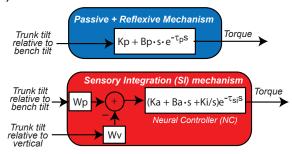




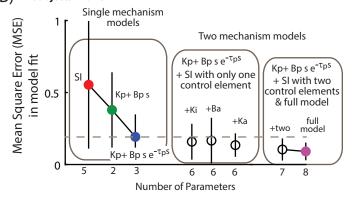




A) Control mechanisms

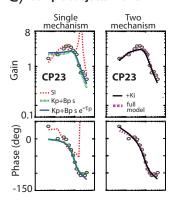


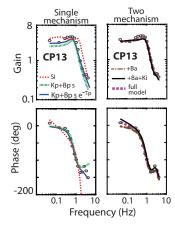
B) Subjects with CP



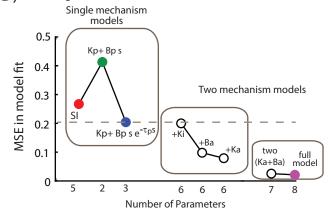
Single mechanism 15 models MSE of full model 10 SI Kp+Bps Two mechanism models 5 +Ki +Ba +Ka +two_ full model Ō Φ Kp+ Bp s e^{-τpS} 0 5 2 3 6 6 **Number of Parameters**

C) Sample subjects with CP

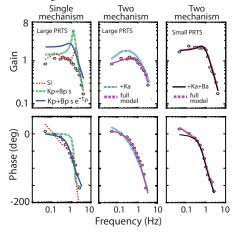


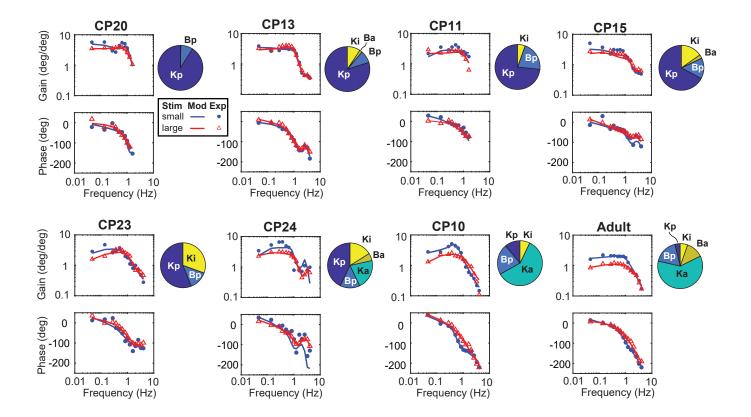


D) Average Adult



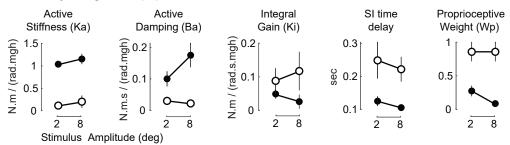
と) Average Adult data





A)

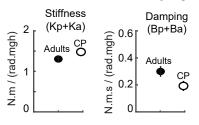
Sensory Integration (SI) mechanism

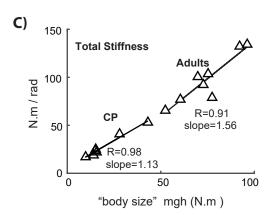


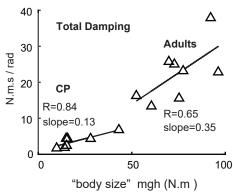
Passive + Reflexive mechanism

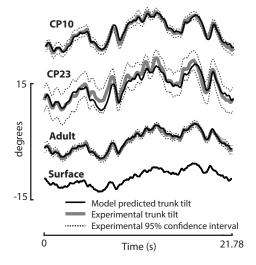
Reflexive Passive Reflexive Stiffness (Kp) Damping (Bp) Time Delay N.m.s / (rad.mgh) N.m / (rad.mgh) 0.08 0.2 0.06 0.18 1 sec 0.16 0.5 0.04 0.14 0 0.02 2 2 Stimulus Amplitude (deg)

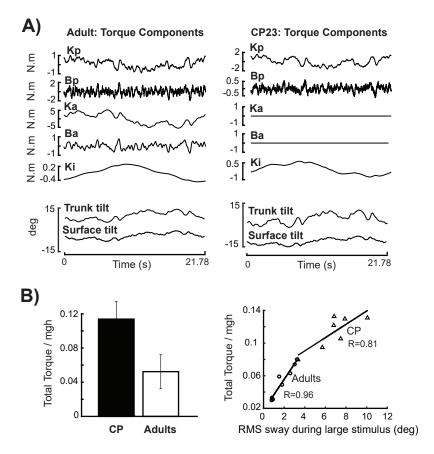
B) Total Stiffness and Damping

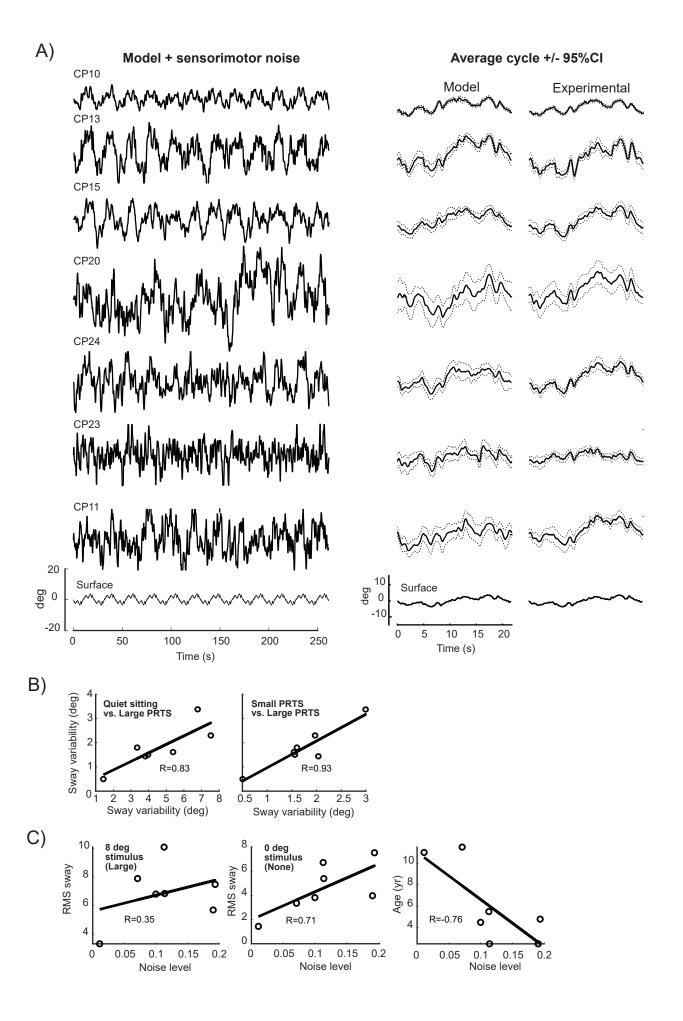


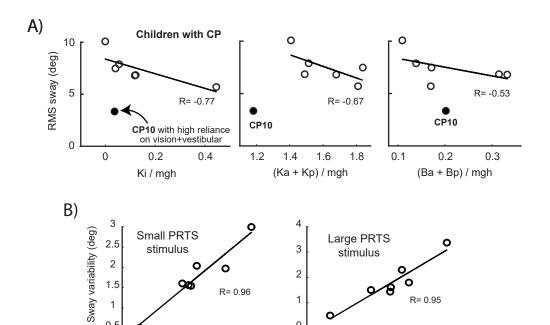












o **l**

3 4 5 6 RMS sway (deg)

4 6 8 RMS sway (deg)

