1	Case Studies in Neuroscience:
2	A Dissociation of Balance and Posture Demonstrated by
3	Camptocormia
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26 Abstract

27 Upright stance in humans requires an intricate exchange between the neural mechanisms that 28 control balance and those that control posture; however, the distinction between these control 29 systems is hard to discern in healthy subjects. By studying balance and postural control of a 30 participant with camptocormia – an involuntary flexion of the trunk during standing that 31 resolves when supine – a divergence between balance and postural control was revealed. A 32 kinematic and kinetic investigation of standing and walking showed a stereotyped flexion of the upper body by almost 80 degrees over a few minutes, and yet the participant's ability to 33 34 control their center of mass within their base of support and to compensate for external 35 perturbations remained intact.

36 This unique case also revealed the involvement of automatic, tonic control of the paraspinal 37 muscles during standing and the effects of attention. Although strength was reduced and MRI 38 showed a reduction in muscle mass, there was sufficient strength to maintain an upright 39 posture under voluntary control and when using *geste antagoniste* maneuvers or "sensory 40 tricks" from visual, auditory and haptic biofeedback. Dual-tasks that either increased or 41 decreased the attention given to postural alignment would decrease, or increase the postural 42 flexion, respectively. The custom-made, 'twister' device that measured axial resistance to 43 slow passive rotation revealed abnormalities in axial muscle tone distribution during 44 standing. The results suggest that the disorder in this case was due to a disruption in the 45 automatic, tonic drive to the postural muscles and myogenic changes were secondary.

46

47 New & Noteworthy

48 By studying an idiopathic camptocormia case with a detailed biomechanical and

49 sensorimotor approach, we have demonstrated unique insights into the neural control of

50 human bipedalism *i*) balance and postural control cannot be considered the same neural

51 process, as there is a stereotyped abnormal flexed posture, without balance deficits,

52 associated with camptocormia, and *ii*) posture during standing is controlled by automatic

53 axial tone but 'sensory tricks' involving sensory biofeedback to direct voluntary attention to

54 postural alignment can override, when required.

55

56 Keywords: Camptocormia, Balance, Posture, Standing, Axial Tone

57 Introduction

58 Terms such as 'postural control' and 'balance' are often used interchangeably in literature on human stance, but equating these control systems is misleading. Upright stance actually 59 60 requires two different actions: *i*. posture, that is maintaining alignment of the body segments 61 with respect to each other and to external references (gravitational vertical, visual vertical, or 62 the support surface), and *ii*. balance, that is the ability to avoid falling in both static (e.g. quiet standing) and dynamic situations (e.g. walking, external perturbations). 63 64 To maintain normal standing, the mechanisms of posture and balance are closely interdependent so it is easy to see why they may be considered a single process. The balance 65 66 system may need to respond to a change of body alignment and conversely, changes in body 67 alignment may occur in anticipation of, or following, a threat to balance. Despite this intimate relationship, the mechanisms by which the central nervous system controls balance and 68 posture during standing may be different so it is unclear whether abnormal control of posture 69 70 necessarily impairs control of balance.

71

72 Camptocormia is a rare condition characterized by an involuntary forward flexion of the 73 trunk that is present in standing and walking, but resolves when supine. Although postural 74 alignment is impaired in this condition, it is unknown to what extent balance is also affected. A previous study has shown that when healthy subjects voluntarily assume a flexed posture 75 76 they are more unstable when perturbed (Jacobs et al. 2005). In the current study, the flexed 77 posture is the preferred posture, so the balance responses may be adapted to this position, or 78 they may show similar impairment. As balance control during quiet stance may not reflect 79 the control during perturbations and movement, different domains of balance control need to 80 be evaluated.

81 The pathogenesis of camptocormia can generally be divided into two camps: a disorder that 82 causes weakness in extensor postural muscles (Laroche et al. 1995; Mahjneh et al. 2002; 83 Margraf et al. 2010) or a central dystonic disorder that disrupts axial postural tone (Reichel et 84 al. 2001; Slawek et al. 2003; Azher and Jankovic 2005; Melamed and Djaldetti 2006). 85 Muscle fibers can be activated through either voluntary motor drive that requires conscious 86 attention to achieve a particular goal, or through automatic tonic drive. Tonic drive is an 87 unconscious process, whereby continuous nerve impulses activate the muscle to keep it in a 88 partially contracted state. When humans stand upright, tonic drive is automatically increased 89 in antigravity muscles to keep them extended in response to the low-intensity stretch from 90 gravity. Voluntary drive has strong cortical projections, whereas muscle tone is derived from 91 a number of subcortical structures, most notably the basal ganglia and brainstem (Takakusaki 92 et al. 2003). Therefore, measuring the force generated from maximal voluntary contractions 93 or muscle imaging does not reveal the extent to which automatic, tonic control of muscles 94 used for postural alignment in stance is disturbed. Quantifying axial postural tone is 95 traditionally problematic, however, our unique "twister" device (Gurfinkel et al. 2006) 96 allowed us to measure, for the first time, automatic axial hip and trunk tone in a 97 camptocormia subject during stance. We also manipulated the relative amount of voluntary 98 versus automatic control to postural alignment with a dual task to divert attention or with 99 sensory biofeedback ('sensory tricks') to add a voluntary postural goal.

100

101 Case history

102 The subject was an active and independent 81 year old female with a twenty-year history of 103 camptocormia. She reported a sudden onset of her flexed posture that occurred while she was 104 hiking. Her posture was severely flexed forward when standing and walking (Figure 1A) but 105 straight when supine (Figure 1C). She maintained an erect posture when using a rolling

106 walker (Figure 1B), but at home, she preferred the flexed posture to avoid using a walker. 107 Ascending a staircase was performed "on all fours". She slept comfortably on either her back 108 or side. She had some intermittent lumbosacral pain but there was no evidence of stroke; no 109 parkinsonian signs of bradykinesia, rigidity, or tremor; no mental status abnormalities and 110 deep tendon reflexes were intact. MRI of the brain was unremarkable. MRI of the lumbar and 111 thoracic spine indicated no structural problems, but the para-spinal (multifidus and 112 longissimus) muscles showed some fatty infiltration (Figure 1D). The iliopsoas muscles by 113 comparison looked normal. Therapeutic strategies had been ineffectual, including; levodopa, 114 trihexphenidyl, baclofen, and botulinum toxin A injections to the rectus abdominis and psoas 115 muscles.

- 116
- 117

[Figure 1]

118 Methods & Results

Signed consent was obtained and the OHSU Institutional review board had approved all
experimental procedures. A healthy subject that matched the camptocormia subject by age,

121 gender, height and weight was used as a control in some conditions.

122

123 Posture versus Balance

The transition from the upright to the flexed posture occurred over 2-3 minutes with a stereotyped time course when measured in 3 sessions over 6 months (exponential time constant of 32±1.2s, Figure 2A). To maintain equilibrium during quiet standing, the nervous system must maintain the vertical projection of the position of the body's center of mass (CoM) over the base of support (the feet). The CoM represents the unique point where the weighted relative position of the distributed mass sums to zero. CoM position and segmental alignment were calculated from 29 reflective markers placed on body landmarks, sampling at

60 Hz (Motion Analysis, Santa Rosa, CA). An inertial sensor placed on the cervical spine, 131 132 sampling at 50 Hz (Xsens, Enschede, The Netherlands) measured trunk tilt with respect to 133 gravity and force plates measured ground reactive forces at 480Hz. 134 135 The flexion did not simply involve the trunk (as implied by the "cormia"/trunk) rather, all 136 axial segments underwent a change in alignment (Figure 2A). Flexion of the trunk and hips 137 was accompanied by extension of the knee and ankle, acting to move the pelvis backward and 138 thus stabilize balance. The overall effect was maintenance of the center of pressure (CoP) and 139 the position of the CoM in the horizontal plane over the base of support (Figure 2B). The 140 back extensor EMG increased as the trunk tilted off vertical and was maintained over the 141 early phase of the forward trunk flexion. However, the muscle activity reduced and became 142 relatively quiet after approximately 40 degrees of trunk flexion (Figure 2C). 143 144 [Figure 2] 145 146 Balance response latencies of the CoP in responding to unexpected forward (mean±S.D. 147 122 ± 9 ms) and backwards (115 ± 12 ms) translations of the standing surface were normal 148 relative to age-matched control subjects (Nashner 1993). In addition, postural sway measured 149 from peak-to-peak displacement of the CoP was normal on all six items of the Neurocom 150 Sensory Organization Test when standing; i) eves open, ii) eves closed, iii) sway referenced 151 to visual field, iv) sway referenced to the standing surface, v) sway referenced to the standing 152 surface without vision, vi) sway referenced to both the visual field and support surface. This 153 demonstrates normal integration of visual, vestibular and proprioceptive input for balance. 154 Also, postural responses elicited by vibrotactile stimulation (80Hz) of the bilateral achilles

tendons for 10 seconds resulted in, a transient forward CoP displacement $(30 \pm 3mm)$,

156 consistent with normal responses (Thompson et al. 2007).

157

To determine whether the deficit in postural alignment was due to a problem with a sense of
verticality, as is often seen in PD (Vaugoyeau et al. 2010) or following brainstem (Yang et al.
2014) or hemispheric stroke (Perennou et al. 2008), the tilt of a hand-held rod with respect to
gravity was recorded while subjects were blindfolded for 3 minutes (Wright and Horak
2007). The rod was held near to vertical (± 1degree) even while the body flexed forward,
indicating a normal sense of verticality.

164

165 Voluntary versus Automatic Postural Control

166 The camptocormia subject had sufficient strength to rise from sitting to standing without the 167 use of her arms. Furthermore, on the experimenter's command, the subject was able to extend 168 her body to the upright (Figure 2C). Maximum isometric strength in the trunk flexors and 169 extensors was measured from a seated position with a dynamometer (Ametek MSC Series) 170 attached with a strap under the arms. The dynamometer was attached to a fixed support in 171 front, and then behind the subject to measure trunk extensor and flexor strength respectively. 172 Three measurements were made with a rest period of 1 minute between each attempt and the 173 average torque was calculated and normalized to body weight and height. Peak isometric 174 trunk extension about the hip joint center was 4.16±0.36Nm and flexion force was 175 4.04±0.19Nm, which was 48% and 77% respectively of normal adult female strength (Keller 176 and Roy 2002).

177

178 <u>"Sensory Tricks"</u>: While standing, the subject performed dual-tasks that either increased or
179 decreased the attention given to postural alignment. Increasing attention to posture was

180 achieved in three ways: i) audio-biofeedback with a tone that increased in pitch and volume 181 with forward sagittal tilt of the trunk measured with an inertial sensor; *ii*) visual biofeedback 182 of the trunk angle in space was visually presented on a 2-D scale on a screen held in front of 183 the subject; *iii*) aiming a laser attached to a toy pistol to a circular target located 3m away 184 from the subject at shoulder height. The subject was given no instruction other than to keep 185 the laser pointed on the target, however, the task was easier with an upright posture. The 186 attention of the subject was diverted away from postural alignment by counting backwards by 187 threes from a random number between 100 and 200.

188

The subject was able to maintain an upright posture for over 3 minutes during both the visual and the auditory biofeedback tasks (Figure 3A). When the dual-task was to aim a laser pistol on a target, there was some forward tilt of the trunk relative to space but the amplitude of the flexion over the same time scale was reduced by half (32° versus 65°). However, when attention was diverted by a serial subtraction dual-task, the rate of trunk bending occurred at a faster rate than quiet standing (time constants 22s versus 32s respectively).

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196 Walking: When the camptocormia subject walked forward, the rate of forward flexion of the 197 trunk was similar to flexion during quiet stance with eyes open (time constant = 36 s) (Figure 198 3Biii). In contrast, during backward and sideways walking, the tilt of the trunk had no 199 significant flexion over 3 minutes. Both backward and sideways walking were associated 200 with backward extension of the arms at the shoulders, the "backswept wing" sign (Margraf et 201 al. 2016), which may have helped to limit forward motion of the trunk. Forward locomotion 202 is suggested to be more automatic, unlike less habitual walking styles that require greater 203 voluntary cortical control (Hackney and Earhart 2010).

204

205

[Figure 3]

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207	Axial tone: To measure axial tone during upright stance on the "twister" device the support
208	surface slowly oscillated in the horizontal yaw plane with a triangle waveform back and
209	forward at 1 deg/s over an amplitude of 5 degrees (Gurfinkel et al. 2006). The resistance to
210	twisting between the lower body and upper body was measured by affixing either the pelvis
211	(to measure hip tone) or the shoulders (to measure both trunk and hip tone) to an earth-fixed-
212	rigid frame. The peak resistance to the rotation was recorded over 5 continuous waveforms
213	and an average was taken. The torsional resistance measured with this technique is a
214	combination of the tone in both axial flexors and extensors.
215	The camptocormia subject had higher hip torque than the age, sex, and BMI matched control
216	subject by 11.4% (1.85 \pm 0.08 versus 1.66 \pm 0.01 Nm), and lower trunk torque by 42%
217	(1.58±0.09 versus 2.71±0.01 Nm). The ratio of the higher trunk tone relative to the hips seen
218	in the healthy subject is consistent with our previous studies of healthy control subjects
219	(Wright et al. 2007). However this ratio of axial tone distribution was markedly disturbed in
220	the camptocormia subject.
221	

222 **Discussion**

This first biomechanical investigation into camptocormia has exposed some important insights into neural control of human standing. Despite severe impairment of postural alignment during standing and forward walking, balance control mechanisms were shown to be intact. This unique case strikingly underscores the fact that "*postural control*" is not *"balance"*.

Previous research on camptocormia has only focused on the terminal, abnormal trunkposition. In this study, we have observed the stereotyped transition from the upright to the

230 flexed posture, with a time constant of 32 seconds. The rate of forward trunk flexion was 231 found to be consistent over three testing sessions months apart. This observation plus the 232 observation that attention to posture influenced the degree of flexion indicates the difficulty 233 with categorizing camptocormia by the angle of flexion (Margraf et al. 2016; 234 Srivanitchapoom and Hallett 2016). The changes in alignment between body segments were 235 highly correlated; with the forward flexion of the trunk and hips counterbalanced by the 236 extension of the knee and ankle joint, acting to move the pelvis backward to maintain the 237 projection of the body CoM over the base of support. Babinski (1899) described similar body 238 compensation during fast voluntary bending in healthy subjects - the 'Babinski synergy' that 239 was absent in people with cerebellar lesions. The dynamic relationship among the axial 240 segments seen in camptocormia is consistent with a functional Babinski muscle synergy. To 241 enact such a balance synergy, the central nervous system must integrate sensory input 242 regarding joint angles with knowledge of segment lengths and mass distributions, which 243 comes from the areas of the brain that store body schema representations (Holmes and 244 Spence 2004).

The subject also had normal balance reactions to visual, vestibular and vibrotactile stimuli, as well as to perturbations of the standing surface. Together, these results demonstrate that the subject had excellent balance control which is substantiated by the fact that she did not report falling and led an active, independent lifestyle.

The pathogenesis of camptocormia is complex and controversial. The subject had reduced muscle mass and relative weakness of the trunk extensors. Based on similar evidence, others have concluded that the primary cause of camptocormia is myogenic (Margraf et al. 2010; Devic et al. 2012; Renard et al. 2012). However, secondary effects associated with reduced use of spinal extensors could produce similar findings. In fact, as normal people age, it is common to see fatty replacement of para-spinal muscles (Haig et al.

2006) and biopsy shows increased pathological abnormalities (Pearce 2005). Furthermore,
prolonged passive stretching of lower back extensor muscles in static flexion may cause
viscoelastic elongation of the muscles which reduces their force generating capacity (Shin et al. 2009).

259 Before a causal link is drawn between loss of muscle mass and primary muscle 260 pathology, biomechanical principles of standing posture must be considered. Upright posture 261 during normal standing is the equilibrium point of competing forces on the body. To maintain 262 upright standing, the tonic control of the dorsal muscles of the trunk and hips must counteract 263 both the static forward moment of gravity and flexor tone. However, only relatively small 264 extensor forces are needed to keep the torso erect. Experimental and biomechanical modeling 265 work (Cholewicki et al. 1997; Kiefer et al. 1998) shows that less than 5% of the trunk 266 extensor MVC is used for upright standing and walking. A slightly larger percentage of the 267 MVC would be required in this camptocormia case, given the absolute MVC is reduced for 268 the same body mass (8.3% of the MVC). This suggests that even though the back extensors 269 showed some weakness, our camptocormia subject still had sufficient strength to maintain 270 upright posture - which she did, but only when posture was under voluntary control with 271 "sensory tricks".

The ability to voluntarily generate muscle contractions that are sufficient for upright body stabilization have been reported in other severe cases of camptocormia (Lin 2004). When our subject performed dual-tasks that directed her attention toward her posture, body alignment could be maintained near to vertical. In contrast, by distracting attention away from her posture, the rate of forward flexion increased compared to quiet standing. If the disorder had a psychogenic origin the rate of flexion would be expected to decrease, not increase, with distraction.

We propose that geste antagoniste maneuvers, or "sensory tricks" over-ride failing tonogenic 279 280 pathways either with voluntary cortical commands, or via augmented sensory inputs to 281 modulate postural muscle tone directly (Franzen et al. 2011). There are many reports of camptocormia responding well to sensory tricks including: wearing a backpack (Gerton et al. 282 283 2010), touching a support (Azher and Jankovic 2005; Shinjo et al. 2008) and walking 284 backward (Van Gerpen and Van Gerpen 2006). We observed an improvement with each of these "tricks" as well as some new ones - auditory and visual biofeedback. These features are 285 286 characteristic of dystonia. The 'twister' device showed the tonic activity of trunk and hip 287 muscles in the camptocormia subject was disturbed compared to normal subjects. The results, 288 considered in light of the physiological principles of standing, suggest a disruption in the 289 automatic, tonic control of axial muscles as the primary cause of our participant's 290 camptocormia.

291

292 Conclusions

"Balance" and *"postural control"* should not be considered the same process and here is a unique example demonstrating why this is so: a woman with idiopathic camptocormia who had disrupted automatic, tonic control of posture during standing, had excellent balance. Her balance synergies during trunk flexion, and responses to mechanical and sensory perturbations, were all functionally normal despite her stereotyped, flexed posture.

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416 Figure Captions

- **Figure 1.** The camptocormia subject's preferred posture (A), walking with a rolling walker (B), lying
- 421 supine (**C**), and MRI (T1 weighted) of the spine with the arrows indicating fatty replacement of the
- 422 para-spinal muscles (**D**).

426

427	Figure 2. Quiet stance immediately following rising from a chair. A. Postural alignment. The upper
428	graph shows the tilt of the body recorded from C3 with respect to gravity on three trials, each over one
429	month apart. The lower graph shows the mean change in sagittal angle of separate body segments.
430	The ankle angle was defined by the knee, ankle and metatarsal markers; the knee angle from the
431	greater trochanter, knee and ankle markers; the trunk from the C3,T10 and PSIS; and the neck angle
432	from the ear, C3 and T10 markers. The pelvis angle was defined by the angle between the ASIS and
433	PSIS vector and the knee to greater trochanter vector. B. Static balance control. CoP (x , y plane) and
434	CoM (x, y plane) remained stable over time, while CoM in the vertical (z) direction moved closer to the
435	ground. C. Para-spinal EMG and trunk tilt. The camptocormia subject was instructed to "stand
436	straight" after 120s. Paraspinal EMG activity was sampled at 480 Hz, amplified at a gain of 5–10K,
437	band-pass filtered from 75–2000 Hz, and full-wave rectified.

Figure 3. Modulating the amount of voluntary control to postural muscles. All graphs show the tilt of
the upper trunk (C3) relative to gravity. A. Erect posture was better controlled during dual-tasks that
required an upright alignment to perform the task (visual/audio biofeedback and a pistol aiming task),
whereas during the counting backwards task postural alignment degraded more rapidly compared
with normal quiet standing. B. Mean (±S.D.) changes to the trunk alignment during walking: (i)
backward, (ii) sideways and (iii) forward.