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BODY BALANCE AND HYPERMOBILITY

Static and dynamic body balance following provocation of the visual and vestibular systems in females with and without joint hypermobility syndrome

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Summary *Objectives:* Joint hypermobility syndrome (JHS) is a heritable disorder of the connective tissue characterized by excessive joint movement, musculoskeletal pain and neurophysiological deficits (i.e. decreased proprioceptive acuity, altered neuromuscular reflexes). Such deficits may affect body balance thus increasing the risk of injury. The present study aimed at examining static and dynamic body balance following challenge of the visual and vestibular systems in individuals with JHS.

Methods: The sample consisted of 21 females with JHS and 20 controls without signs of JHS. Static body balance was assessed by the degree of anteroposterior and mediolateral deviation of the center of pressure, during 20-sec single-leg stances with eyes opened (EO), eyes closed (EC) and eyes opened with head extension (EO-HE) using a foot pressure platform. Dynamic

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body balance was assessed by the number of landing and balance errors committed during a multiple single-leg-hop-stabilization test.

Results: Nonparametric analysis showed that the JHS-group demonstrated significantly greater (a) mediolateral deviation during single-leg-stance with EO ($p < 0.01$), (b) mediolateral and anteroposterior deviation during single-leg-stance with EO-HE ($p < 0.05$), and (c) number of landing errors ($p < 0.05$) compared to the control group.

Conclusions: Poor static balance following challenge of the vestibular system may be justified by vestibular deficiency and/or insufficient proprioceptive capabilities of the neck. Impairments of dynamic balance in individuals with JHS may be attributed to proprioceptive deficits, which can alter feedforward and feedback mechanisms.

Introduction

Joint hypermobility syndrome (JHS) is an insufficiently understood and often poorly managed multi-system hereditary disorder of the connective tissue (Simmonds and Keer, 2007) that prevails among young, non-Caucasian females (Grahame and Hakim, 2004; Forleo et al., 1993). It is characterized by excessive joint laxity combined with widespread musculoskeletal pain and/or other extra-articular features of connective tissue laxity (revised Brighton criteria) (Grahame et al., 2000), in the absence of signs of infectious, inflammatory and autoimmune disorders (Everman and Robin, 1998). The revised Brighton criteria is a validated (Grahame et al., 2000) and reproducible set of criteria ($k > 0.73$, Juul-Kristensen et al., 2007) that have been commonly used in the diagnosis of JHS. Based on these criteria, an individual is diagnosed with JHS in the presence of (i) a Beighton score of $\geq 4/9$ – a nine-point score that is being gained if someone can passively dorsiflex the little fingers beyond 90° (2 points), passively oppose the thumbs to the flexor aspect of the forearms (2 points), hyperextend the elbows beyond 10° (2 points), hyperextend the knees beyond 10° (2 points), forward flex the trunk with knees straight and the palms of the hands rest easily on the floor (1 point) – and (ii) arthralgia for ≥ 3 months in ≥ 4 joints (major criteria). Alternatively, an individual with JHS may fulfill one of the aforementioned major criteria and at least two minor criteria or none of the major and at least four minor criteria. The minor criteria include (i) a Beighton score of 1–3/9 (or 0–3/9 if aged >50 yrs), (ii) arthralgia for ≥ 3 months in 1–3 joints, back pain for ≥ 3 months, spondylosis or spondylolysis/spondylolisthesis, (iii) acute or recurrent dislocation/subluxation in ≥ 1 joint, (iv) ≥ 3 soft tissue lesions (i.e. epicondylitis, bursitis), (v) Marfanoid habitus (ectomorphic somatotype), (vi) skin abnormalities (i.e. thin and hyperextensible skin, abnormal skin striae), (vii) eye pathology (i.e. drooping eyelids, myopia) and (viii) varicose veins or hernia or uterine/rectal prolapse.

Individuals with JHS have also demonstrated poor proprioceptive capabilities with regard to the knee joint (Baskent et al., 2008; Ferrell et al., 2004; Hall et al., 1995) and the proximal interphalangeal joints (Mallik et al., 1994). Other authors revealed that the reflex activation of certain muscles (e.g. quadriceps) that is usually elicited following stimulation of peripheral nerves (e.g. common peroneal nerve) was either insufficient or absent in this group of individuals suggesting decreased neuromuscular co-ordination (Ferrell et al., 2007).

Another somatosensory-depended function of the human body that has been assessed in individuals with JHS was body balance. Body balance is referred to as the ability to maintain the body's center of gravity over its base of support with minimal sway or maximal steadiness (Horak, 1987; Shumway-Cook et al., 1988). Although it is considered vital for optimal performance of the human body and injury prevention, particularly of the lower limbs (Hrysomallis, 2007), in the JHS population has been investigated only under static conditions (bipedal and unipedal upright stance) with eyes open (Ferrell et al., 2004; Mebes et al., 2008). However, body balance may be examined either under static or dynamic conditions. When static body balance is performed with eyes closed, clinicians may extract additional information of how it is controlled under the interactive information of the somatosensory and the visual systems via the CNS (Gatev et al., 1999). Backward tilting of the head (neck extension) during upright stance may also provide information of the ability of the somatosensory system to compensate for the increasing demands in maintaining body balance due to proprioceptive and vestibular provocation (Brandt et al., 1981). The neck flexors, which are muscles with a high density of muscle spindles (e.g. longus colli), are stretched in this position and may possibly affect postural control by compromising modulation of musculotendinous and capsuloligamentous reflexes (Boyd-Clark et al., 2002). Furthermore postural control, in this position, may be compromised by dysfunction of the vestibular system and especially the otoliths which contain several fluid-filled membranous sensory end-organs. The macula, which is the sensory area of otoliths, support hair cells that act as mechanoreceptors. When the head is tilting backwards, the mass of statoconium membrane prevents these hair cells from returning to their resting position, thus generating tonic signals that represent head position in relation to gravity (Keshner and Cohen, 1989).

Dynamic body balance testing [i.e. the multiple single-leg-hop stabilization test (Riemann et al., 1999)], on the other hand, may provide information with regard to the effectiveness of the feedforward and feedback mechanisms in correcting postural deviations that are necessary in achieving a successful performance during daily and sporting activities. Such testing may also be more appropriate in revealing balance deficiencies particularly in a young population with increased physical activity.

The aim of the present study was to investigate by means of postural sway, the contribution of proprioceptive information generated with and without challenging the visual and vestibular systems in maintaining body balance under static and dynamic conditions in individuals with and without JHS.

Methods

Subjects

The sample consisted of 41 Caucasian female students with JHS ($n = 21$) and without JHS ($n = 20$). All participants were recruited from the Department of Physical Education and Sports Science of a major local University. The revised Brighton criteria were used for the diagnosis of the JHS (Grahame et al., 2000). All subjects who were assigned to the JHS group had $>4/9$ score in Beighton scale as a major criterion and at least two minor criteria (e.g. back pain, joint dislocations, myopia, skin hyperextensibility, arthralgia in one to three joints). The subjects were free of any lower limb or spine pathology, visual, vestibular or balance disorders, other connective tissue disorders (e.g. Marfan's syndrome) and previous surgeries. They also did not participate in organized sports or perform intensive motor activities (working or recreational) of everyday life (Baecke et al., 1982). Information with regard to the past/present medical health was selected prior to the investigation. Before testing each participant signed an informed consent form that had been approved by the University's ethics committee.

Static and dynamic balance testing

Static balance on the dominant leg was examined by means of 20-sec single-leg-stance sways with eyes open (EO), eyes closed (EC) and eyes open – head extended (EO-HE), according to procedures described by Hansson et al. (2010) (Fig. 1A,B). Anteroposterior and mediolateral postural sway was assessed by the vertical (y-component of foot pressure vector) and horizontal (x-component of foot pressure vector) deviation of the center of foot pressure, using a foot pressure distribution platform (FDMS, Zebris Co., Medical GmbH, Germany). The platform was consisted of 1792 capacitive force sensors arranged in a 32 × 56-cm matrix and it was synchronized to a personal computer. Foot pressure signals were recorded at a sampling rate of 120-Hz and analyzed with the WinFDMS software (WinFDMS 0.1× for Windows, zebris Medical GmbH).

Dynamic balance was tested with multiple single-leg-hops (modified Bass test). The participants were required to hop in lateral and diagonal directions and land on 10 prearranged points that were marked on a 3.80-m long canvas mat with 2.5-cm^2 fluorescent yellow-tapes. Prior to testing each participant was informed about the testing procedures and the scoring system, and performed a 10-min warm-up consisted of 5-min pedaling on a cycle ergometer, 5-min stretching exercises for the lower limbs and one or two trials of the actual test for familiarization. The test began with each participant standing at the starting point on the foot to be tested, facing forward and with the hands firmed on the waist (Fig. 1C). Before each hop the participant was allowed to look at the next point and land on or as close as possible to it (landing phase) trying to remain for 5-sec in a single-leg-stance position (balancing phase) (Riemann et al., 1999). The errors committed during the landing and balancing phases were recorded by the tester using a video-camera (Microsoft LifeCam, VX 7000, Model 1121, Microsoft Corporation, USA) and calculated based on the Balance Error Scoring System [ICC (2,1): 0.70–0.92] (Riemann et al., 1999). On landing, according to the Balance Error Scoring System, each participant should (i) cover the mark, (ii) avoid stumbling, (iii) remain with the hands on the iliac crests and (iv) the landing foot should point straight ahead (landing errors). During the 5-sec balance period, the participant should maintain a stable position (i) with the hands on the iliac crests (ii) without touching down or (iii) touching the contralateral limb and (iv) avoid excessive (>30°) flexion, abduction and extension of the contralateral limb (balance errors). Static and dynamic balance testing was conducted in a randomized order.

Statistical analysis

Initial diagnostics on the two static balance measures (horizontal and vertical deviation) revealed that both dependent variables demonstrated lack of homogeneity of variance (Levene's test) and severe non-normality (boxplots, histograms, the Shapiro–Wilk test). Therefore, nonparametric ANOVAs for repeated measures (Friedman tests) were used to examine potential differences between the three experimental conditions (EO, EC, EO-HE), whereas the two groups of subjects (JHS, controls) were compared using non-parametric Mann–Whitney tests. All post-hoc analyses for the between conditions differences were corrected for inflation of type I error rate due to multiple comparisons using the non-parametric Wilcoxon tests with Bonferroni correction.

Differences between the two groups (JHS, controls) in terms of the errors committed during the landing and balancing phases in the dynamic balance testing were examined using independent t-tests. All statistical analyses were carried-out using SPSS 17.0. Statistical significance was tested at $\alpha = 0.05$ probability level of type I error rate.

Results

The differences between the two groups with regard to the anthropometric characteristics (body weight, body height and BMI) and the physical activity level were not statistically significant (Table 1).

Based on Brighton criteria the participants in the JHS group presented one major (Beighton score $\geq 4/9$) and $\geq 2/7$ minor criteria. Beighton's score was significantly greater in the JHS group compared to the control group (6.6 ± 1.5 vs. 2.1 ± 1.7, $p < 0.001$). The majority of the participants in the JHS group presented 2/7 minor criteria ($n = 18$). Three more participants in the same group presented 3/7 minor criteria. The criteria met by these individuals were skin hyperextensibility ($n = 15$), low back pain ($n = 10$), arthralgia in one to three joints ($n = 7$), joint dislocations ($n = 5$), myopia ($n = 4$) and Marfanoid habitus ($n = 4$). The participants in the control group presented similar criteria; however the total number of criteria that were met by each one of these individuals was less than two.

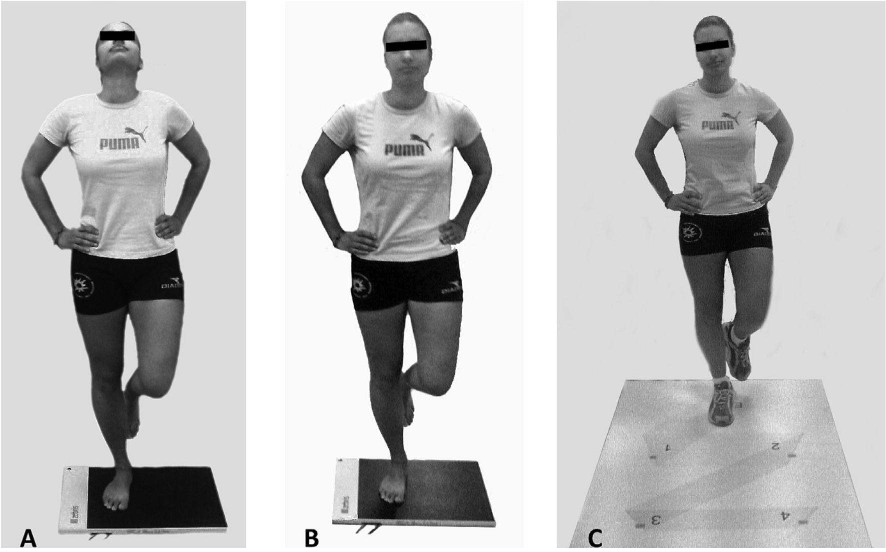

Figure 1 Single leg stance with eyes open (A) and eyes open and head extension (B) for static balance testing. Starting position of multiple-single-leg-hop stabilization test for dynamic balance testing (C).

Horizontal deviation (mediolateral sway) was significantly greater during single-leg-stance with EO ($p < 0.01$) and EO-HE ($p < 0.05$) in the JHS group compared to the control group (Fig. 2A). The JHS group presented also greater vertical deviation (antero-posterior sway) compared to the control group. However the differences between the groups were statistically significant only during single-leg-stance with EO-HE ($p < 0.001$) (Fig. 2B).

Both JHS and control group demonstrated significantly greater mediolateral and antero-posterior postural sways during single-leg-stance with EC compared to single-leg-stance with EO-HE and EO ($p < 0.001$). The single-leg-stance with EO-HE was significantly different compared to EO only for antero-posterior sway in the JHS group ($p < 0.001$).

With regard to the dynamic balance the analysis revealed a greater number of landing and balance errors in the JHS group compared to the control group; however the differences were statistically significant only for landing errors ($p < 0.05$) (Fig. 3).

Discussion

The findings of the present study revealed both static and dynamic balance impairments in females with JHS compared to individuals without JHS. The most significant changes were observed during one-leg stance with eyes open and the head extended (EO-HE), a position where postural sway could be challenged by deficiencies of the vestibular system (Brandt et al., 1981). Although there is no data suggesting deficiency of the structure (labyrinth and otoliths) and function of the vestibular system in individuals with JHS there is indirect evidence that supports otherwise (Aoki et al., 2012; Gazit et al., 2003). It has been shown that vestibular disorders, due to dysfunction of the otolith organs, may provoke orthostatic hypotension (Aoki et al., 2012). Otolith stimulation, which can be generated by off-vertical axis rotation (e.g. head extension), may alter the discharges of vasoconstriction efferent, affecting regulation of blood pressure during movement or changes in posture (Aoki et al., 2012; Yates, 1992). This autonomic dysfunction has been also observed in individuals with JHS (Gazit et al., 2003) and despite the fact that the vestibular system was not examined in the present study its potential dysfunction might affected postural control.

Table 1 Anthropometric characteristics and level of physical activity of the participants in the group with JHS ($n = 21$) and control group ($n = 20$).

Anthropometric characteristics	JHS group	Control group
Age	21.7 (1.7)	21.5 (1.7)
Height (cm)	165.2 (7.1)	166.1 (6.5)
Body weight (kg)	58.8 (8.7)	57.3 (5.6)
BMI (kg/m^2)	21.5 (2.5)	20.8 (1.8)
Physical activity (0–15 points)	9.4 (1.4)	9.0 (1.4)

Alternatively, if the structure and function of the vestibular system is normal, the postural disturbance observed during one-leg stance could be justified by deficits in the somatosensory system, provided that the visual input is intact (Yasuda et al., 1999). Normally the successful control of static posture, when it is challenged, requires intact proprioceptive information, particularly from the neck and ankle, in order to provide a continuous update of the internal representation of stance and to trigger pre-programmed responses (Gatev et al., 1999; Keshner and Cohen, 1989). If the afferent information from the proprioceptive receptors that are located within passive (i.e. ligaments, capsule) and active (muscles) stabilizers is insufficient the timing and the sequence of muscle activation can be modulated (Solomonow et al., 1998) affecting body sway. Females with JHS, in the present study, demonstrated a significant increase in horizontal deviation during one-leg stance with EO and EO-HE. The increased horizontal static postural deviation that has been demonstrated by male and/or female patients with JHS

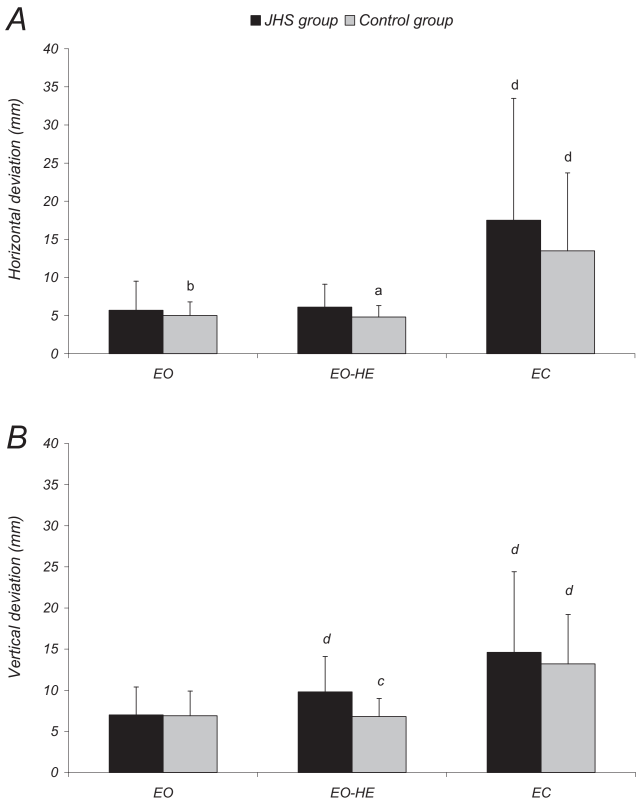

Figure 2 Medians and interquartile ranges of horizontal (A) and vertical deviations (B) of postural sway that recorded by the JHS and control groups during single-leg-stance with eyes open (EO), eyes open and head extension (EO-HE) and eyes closed (EC). ^{a} $p < 0.05$, ^{b} $p < 0.01$ and ^{c} $p < 0.001$ for differences between groups in each stance; ^{d} $p < 0.001$ for differences between pair of stances in each group.

during single-leg-stance with EO in a previous study (Mebes et al., 2008), was attributed to the poor proprioceptive signaling from the receptors of the knee joints (Baskent et al., 2008; Ferrell et al., 2004; Hall et al., 1995). Poor proprioceptive signaling, however, could also be derived from receptors located in more cephalad body parts, as joint hypermobility is a feature that has a global effect in individuals with JHS. Longus colli for example, which is a flexor of the neck with a high density of muscle spindles, may play an important role in proprioceptive information and hence postural control, particularly when it is stretched during stance (bipedal or unipedal) with head extension (Boyd-Clark et al., 2002).

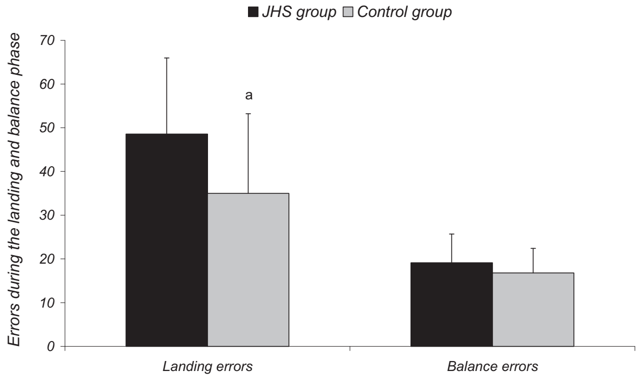

Figure 3 Means and standard deviations of the errors committed by the JHS and control group during the landing and balancing phase of the multiple-single-leg-hop stabilization test. ^{a} $p < 0.05$ between groups.

In addition to the proprioceptive deficits, the impaired postural control that was observed during dynamic balance testing could be attributed to differences in feedforward and feedback mechanisms. The feedforward mechanism is an unconscious function that is regulated by the CNS and occurs prior to the initiation of a movement by stabilizing individual body parts for optimal performance (Page, 2006). This automatic level of processing is essential in protecting joints during different motor activities through pre-activation of muscles that are important in maintaining postural and joint stability (Hodges and Richardson, 1997a, 1997b). The greater number of landing errors that committed by the JHS group compared to the control group, during the single-leg-hop-test, could be justified based on differences in the aforementioned mechanism. Chappell et al. (2007) recorded an increased EMG activity of the quadriceps and hamstrings 50 ms before landing that follow a vertical stop-jump, suggesting that lower extremity motion patterns were pre-programmed during this task. These anticipatory postural adjustments are considered necessary in order to maintain optimal postural control during the performance of a functional task such as jumping.

The increased number of balance errors is an indication of poor compensatory reactions of the distally located body segments. Compensatory reactions are more likely to be related with insufficient function of the feedback mechanism. The ability of maintaining balance for 5-sec in each point during single-leg-hop testing is primarily based on the modulation of the lower limb muscles' reflex activity following the integrated information sent to the CNS by the joint and muscle proprioceptors. Abnormal reflexes have been observed in females with JHS by Ferrell et al. (2007). By stimulating group I afferent fibers of the common peroneal nerve, the researchers in this study were unable to elicit quadriceps' reflex activation in half of the participants with JHS that they examined. The absence of such reflexes may compromise knee joint stability justifying in part the lack of balance that observed following landing after a single-leg-hop in the present study.

Our findings suggest that both static and dynamic body balance testing may be an important tool in screening as well as in longitudinal evaluation (e.g. before and after an exercise intervention program) of the somatosensory status in JHS patients. Furthermore clinicians are advised to incorporate in rehabilitation programs body balance training, including exercises that mainly challenge the somatosensory and vestibular systems, in order to improve functional performance and to prevent injury or falls, particularly in older adults (Rombaut et al., 2011). This recommendation is supported also by previous studies which have shown that balance training was beneficial in individuals with JHS with regard to symptom's relief and motor performance improvements (Ferrell et al., 2004).

In conclusion, the impaired static and dynamic body balance in females with JHS that was revealed in the present study may be attributed to poor proprioceptive capabilities and/or vestibular dysfunction and the subsequent modifications of the feedforward and feedback mechanisms. However, our findings should be viewed in light of the limitations that related to sample selection such as the age, health status and neuromuscular coordination level (as suggested by the reported physical activities). Body balance and the potential impact on postural sway could be different if the participants were older (Prado et al., 2007), with health problems or injuries (e.g. osteoarthritis, musculoskeletal injuries) (Hassan et al., 2001) and/or different levels of neuromuscular coordination (e.g. gymnasts) (Asseman et al., 2008).

Further research is required to investigate the subsystems (e.g. proprioceptive system, vestibular system) that are involved in body balance performance in individuals with JHS of different age, health status and physical condition.

Conflict of interest

The authors have declared no conflicts of interest.

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