Clinical Biomechanics 28 (2013) 318–324

Contents lists available at SciVerse ScienceDirect

Clinical Biomechanics

journal homepage: www.elsevier.com/locate/clinbiomech

Dynamic balance during gait in children and adults with Generalized Joint Hypermobility

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ARTICLE INFO

Article history:
Received 28 August 2012
Accepted 9 January 2013

Keywords:
Joint hypermobility
Locomotor balance
Dynamic balance
Angular dispersion
Anchoring index
Walking
Gait

ABSTRACT

Background: The purpose of the study was to investigate if differences of the head and trunk stability and stabilization strategies exist between subjects classified with Generalized Joint Hypermobility and healthy controls during gait. It was hypothesized that joint hypermobility could lead to decreased head and trunk stability and a head stabilization strategy similar to what have been observed in individuals with decreased locomotor performance.

Methods: A comparative study design was used wherein 19 hypermobile children were compared to 19 control children, and 18 hypermobile adults were compared to 18 control adults. The subjects were tested during normal walking and walking on a line. Kinematics of head, shoulder, spine and pelvis rotations were measured by five digital video cameras in order to assess the segmental stability (angular dispersion) and stabilization strategies (anchoring index) in two rotational components: roll and yaw.

Findings: Hypermobile children and adults showed decreased lateral trunk stability in both walking conditions. In hypermobile children, it was accompanied with decreased head stability as the head was stabilized by the inferior segment when walking on a line. Several additional differences were observed in stability and stabilization strategies for both children and adults.

Interpretation: Stability of the trunk was decreased in hypermobile children and adults. This may be a consequence of decreased stability of the head. Hypermobile children showed a different mode of head stabilization during more demanding locomotor conditions indicating delayed locomotor development.

The findings reflect that Generalized Joint Hypermobility probably include motor control deficits.

1. Introduction

Generalized Joint Hypermobility (GJH) is defined as a condition in which joint range of motion (ROM) is increased compared to the general population when age, gender and ethnicity are taken into consideration (Hakim and Grahame, 2003; Remvig et al., 2007; Simmonds and Keer, 2007). GJH can either be acquired through excessive stretching in sports which requires flexibility to a high degree (Gannon and Bird, 1999), or it can be inherited (Dalgleish, 1997; Grahame, 1999).

GJH is often associated with pain, musculoskeletal and soft tissue complaints, such as osteo-arthritis, arthralgia, frequent luxations and subluxations (Acasuso et al., 1993; Hudson et al., 1998; Scott et al., 1979).

Some of these conditions are included in the criterion set for Benign Joint Hypermobility Syndrome (BJHS) (Grahame et al., 2000), a disorder with unknown pathophysiology (Remvig et al., 2007; Simmonds and Keer, 2007). BJHS may be identical to Ehlers–Danlos Syndrome (EDS), hypermobile type (Grahame, 1999; Tinkle et al., 2009), and as such be part of the Heritable Connective Tissue Disorders (HCTD), which also includes other syndromes with GJH such as Marfan's Syndrome and Osteogenesis Imperfecta. In adults, GJH as defined in the BJHS criterion set (e.g. Beighton score of 4 tests positive out of 9, GJH4) has also been reported to be present together with impaired physical function, such as decreased proprioception (Hall et al., 1995; Sahin et al., 2008a), decreased knee muscle strength (Sahin et al., 2008b) and reduced postural balance (Mebes et al., 2008). It is unknown whether or not GJH affects dynamic balance, but the impaired physical function in subjects with GJH may likely affect the dynamic balance.

Dynamic balance is a complex task, but a necessity for human gait, because it maintains the body in equilibrium during the propulsion of the body, which involves highly destabilizing forces (Assaiante, 1998). In bipeds, the difficulty of maintaining equilibrium is further accentuated by the fact that the body is only supported by one leg

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http://dx.doi.org/10.1016/j.clinbiomech.2013.01.006

during the swing phase of gait (Assaiante, 1998; Assaiante and Amblard, 1995).

Head stabilization in space is known to be important while walking, because the head houses the visual and vestibular systems which are responsible for detecting loss of balance and control lateral trunk equilibrium (Assaiante and Amblard, 1993). Also, human neck muscles were reported to contain the greatest abundance of muscle spindles, whereas the least was in muscles of the shoulder girdle (Banks, 2006). The head stabilization in space provides clear vision and better vestibular processing and the head hereby acts as a stable reference, on which the stability of the trunk is organized (Assaiante and Amblard, 1995). Because human walking is cyclic it induces oscillations in the trunk and head (Grossman et al., 1988) which thus have to be stabilized (Assaiante and Amblard, 1993; Borel et al., 2002; Grossman et al., 1989; Nadeau et al., 2003; Pozzo et al., 1990). Accordingly, the purpose of the present study was to investigate if differences in the stability and stabilization strategies of the head and trunk exist between individuals with GJH and healthy controls. The fundamental hypothesis was that joint hypermobility would influence the dynamic balance negatively by decreased head and trunk stability and a head stabilization strategy, in which the head is stabilized by the inferior segment, similar to individuals with decreased locomotor performance such as healthy children under the years of seven and in patients with Parkinson's disease (Assaiante, 1998; Assaiante and Amblard, 1993; Mesure et al., 1999).

2. Methods

2.1. Subjects

A comparative study design was used, including children (randomly selected) and adults selected due to their parenting of the children. Totally, 85% of the adults were parents to these children. The children were part of the *Copenhagen Hypermobility Cohor*t (COHYPCO) from two medium size Danish municipalities with approximately 46,750 and 40,215 inhabitants, respectively (Juul-Kristensen et al., 2009; Remvig et al., 2011).

The subjects were clinically diagnosed by two rheumatologists with many years of clinical experience who strictly followed the procedures of the Beighton score, which has shown high reproducibility (Juul-Kristensen et al., 2007). In total 74 subjects were included and they were clinically classified as having Generalized Joint Hypermobility (GJH) or Non-Generalized Joint Hypermobility (NGJH). GJH children were compared with NGJH children, and GJH adults were compared to NGJH adults. There were 19 subjects in each group of children and 18 in each group of adults (Table 1).

Inclusion criteria for children with GJH were a Beighton score of $\geq 5/9$, and a Beighton score of $\geq 4/9$ for adults with GJH. In addition, both groups should fulfill two remaining inclusion criteria: at least one hypermobile knee, i.e. > 10° hyperextension, and no knee arthralgia (children).

NGJH children were classified by a Beighton score of <5 and NGJH adults by a Beighton score of <4. Subjects who had hereditary diseases like Ehlers–Danlos Syndrome, Marfan Syndrome or Osteogenesis Imperfecta, a Body Mass Index (BMI) > 25 or an inability to understand Danish were excluded from both groups.

Table 1
Four groups of subjects and their mean age. Range is specified in brackets.

	Number	Age (years)
Children GJH	19	10.12; [9–11]
Children NGJH	19	10.16; [10–11]
Adults GJH	18	39.64; [32–51]
Adults NGJH	18	40.09; [31–47]

All patients and subjects gave their written informed consent to the experimental procedures, which were approved by the local ethics committee. There were no conflicts of interests.

2.2. Experimental procedure

The experiments were conducted in a 10 m long gait lab with five digital video cameras (Canon MW600) operating at 50 frames per second. Three recessed force platforms (AMTI OR6-5-1) were used to determine a full gait cycle. Cameras and force platforms recorded the gait data in synchrony.

The subjects were taught to walk at 4.5 km/h ± 10% by immediate feedback on walking speed (normal walking), which was measured by photocells. The subjects were exposed to two different walking conditions: 1) normal walking and 2) walking on a white line marked on the floor across the force plates. During the latter condition the subjects were allowed to walk at self-selected walking speed. For each condition three gait cycles were selected for further analysis.

Twenty reflective markers were placed symmetrically on the following anatomical landmarks: The fifth metatarsal joint, the calcaneus, the lateral malleolus, the tibial tuberosity, the lateral femoral epicondyle, the greater trochanter, the anterior superior iliac spine, the posterior superior iliac spine, acromion and the mastoid process. In addition, five unpaired reflective markers were placed on: sacrum, three vertebral processes (C_7, T_6, L_2) and the top of the head.

This marker arrangement was used to measure the horizontal (yaw) and lateral (roll) rotations of the head, shoulders, spine, and pelvis. The marker setup was a slight modification of Mallau et al. (2007). The head segment was defined by the markers placed on the left and right mastoids. The shoulder segment was defined by the markers on the left and right acromion and the pelvis was defined by markers placed on the left and right greater trochanter, respectively.

The spine was subdivided into three segments in order to analyze the movements within the spine. The lumbar part was located between sacrum and L_2, the thoracic part between L_2 and T_6, and the cervical part between T_6 and C_7. The arrangement of the markers is shown in Fig. 1.

2.3. Data treatment and calculations

The video recordings were digitized and stored on a PC. Three dimensional coordinates were reconstructed by direct linear transformation using the Ariel Performance Analysis System (APAS, Ariel Dynamics Inc., San Diego, CA, USA). Accuracy and reproducibility of the APAS system have been evaluated by Richards (1998). All coordinates were low-pass filtered at 6 Hz by a 4th order Butterworth filter. Each of the obtained gait cycles was time normalized to 100% gait cycle by linear interpolation in MATLAB (version 2011a).

2.4. Control parameters

The filtered coordinates were input to software written in MATLAB, which calculated two control parameters expressing stability (Angular Dispersion) and stabilization strategies (Anchoring Index) of the previously defined segments.

The absolute angles (with respect to external axes) were calculated, and for each trial the standard deviation of the absolute angular distributions (SD_{abs}) was calculated in order to obtain the Angular Dispersion (AD).

$$AD = SD_{abs}.$$

The AD assessed the amplitude of the oscillations in the segments. If AD of a given segment was less than AD of an inferior segment, the oscillations were attenuated from one segment to the next (Mallau et al., 2007).

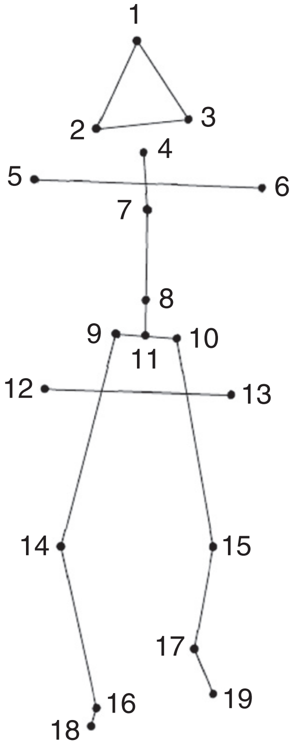

Fig. 1. Arrangement of some of the 25 markers: The top of the head (1), the mastoid processes (2, 3), the acromion processes (5, 6), the vertebral processes of C7 (4), T6 (7) and L2 (8), the posterior superior iliac spine (9, 10), the greater trochanters (12, 13), the lateral femoral epicondyles (14, 15); the lateral malleolus (16, 17), the fifth metatarsal joints (18, 19) and the sacrum (11).

Anchoring Index (AI) was used to compare the stabilization with respect to both external space and the inferior anatomical segment (Mallau et al., 2007). It was calculated according to the formula:

$$\mathrm{AI} = \frac{(\mathrm{SD_{rel}})^2 - (\mathrm{SD_{abs}})^2}{(\mathrm{SD_{rel}})^2 + (\mathrm{SD_{abs}})^2}.$$

$\mathrm{SD_{abs}}$ is the standard deviation of the absolute angular distributions and $\mathrm{SD_{rel}}$ is the standard deviation of the relative angular distribution of the considered segment relative to the inferior segment.

AI indicates the dependency between two consecutive segmental movements. The values of AI vary between −1 and 1, and a positive AI value for a given segment indicates that stabilization occurs in space rather than upon the inferior segment. A negative AI value for a given segment indicates stabilization upon the inferior segment rather than in space (Mallau et al., 2007; Mesure et al., 1999; Nadeau et al., 2003). It should be noted that a given segment can be stabilized in space despite of a negative AI. This is the case, when the inferior segment is stabilized in space.

2.5. Statistics

The results are presented as group mean values and (SD). Data was tested to be normally distributed by the Kolmogorov Smirnov z-test and between group differences in demographic and self-repeated variables were tested by a Students *t*-test (two-tailed). Differences between groups of subjects were tested by a "general linear mixed regression model" with anchoring index and angular dispersion input (one at a time) as dependent variables. Subject was a random factor and trial number was used as repeated factor. The level of significance was set to $P<0.05$. All statistical analyses were performed by the "Statistical Package for Social Sciences" (SPSS, version 18.0.0, IBM, USA).

3. Results

3.1. Children

3.1.1. Roll

During normal walking no significant differences between GJH and NGJH children were observed regarding the roll AD of head, shoulder and hip. However, children with GJH had significantly larger head roll AD than NGJH children during walking on a line ($P=0.042$) (left part of Fig. 2).

During normal walking, the AD decreased significantly from the hips to the shoulders in both groups (NGJH: $P<0.001$; GJH: $P=0.0028$) (left part of Fig. 2) but without further decrease from shoulder to head. This pattern was not present when walking on a line.

In addition to the larger head roll AD when walking on a line, GJH children showed greater AD of the thoracic trunk (normal walking: $P=0.035$; walking on a line: $P=0.029$) and lumbar trunk (normal walking: $P=0.040$; walking on a line: $P=0.030$) in both walking conditions (right part of Fig. 2).

During normal walking no significant difference was observed with respect to the head roll AI whereas during walking on a line GJH children had a significant different roll AI of the head ($P=0.014$). In NGJH children independent head stabilization in space was observed while the GJH children stabilized their head with respect to the trunk, which was reflected by a positive AI in NGJH and a negative AI in GJH (left part of Fig. 3).

During normal walking GJH children had a significantly lower roll AI of the cervical trunk ($P=0.005$) indicating decreased independent stabilization of the upper trunk compared with NGJH children (right part of Fig. 3). This difference was not observed during walking on a

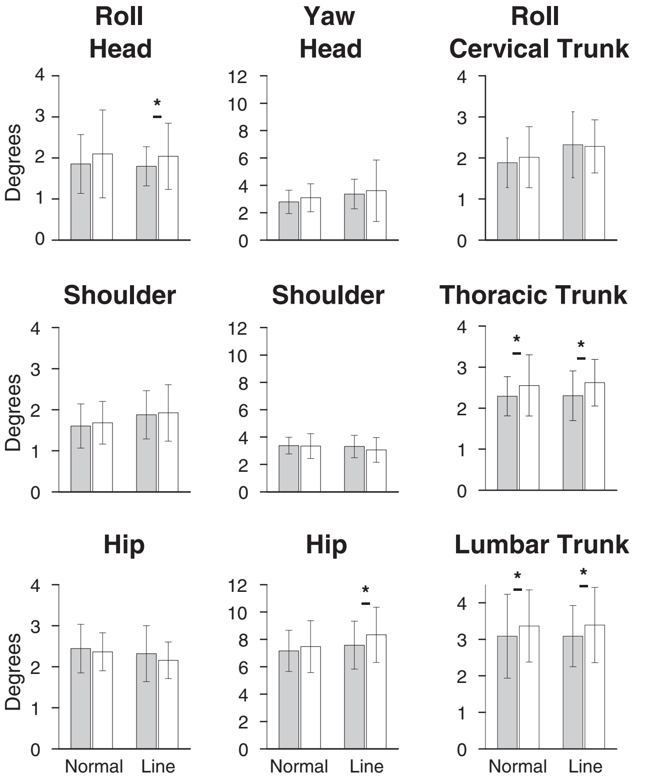

Fig. 2. Mean (SD) of roll and yaw angular dispersions in degrees of the various segments in NGJH (gray) and GJH (white) children in both walking conditions (normal and line). Asterisks indicate statistically significant differences.

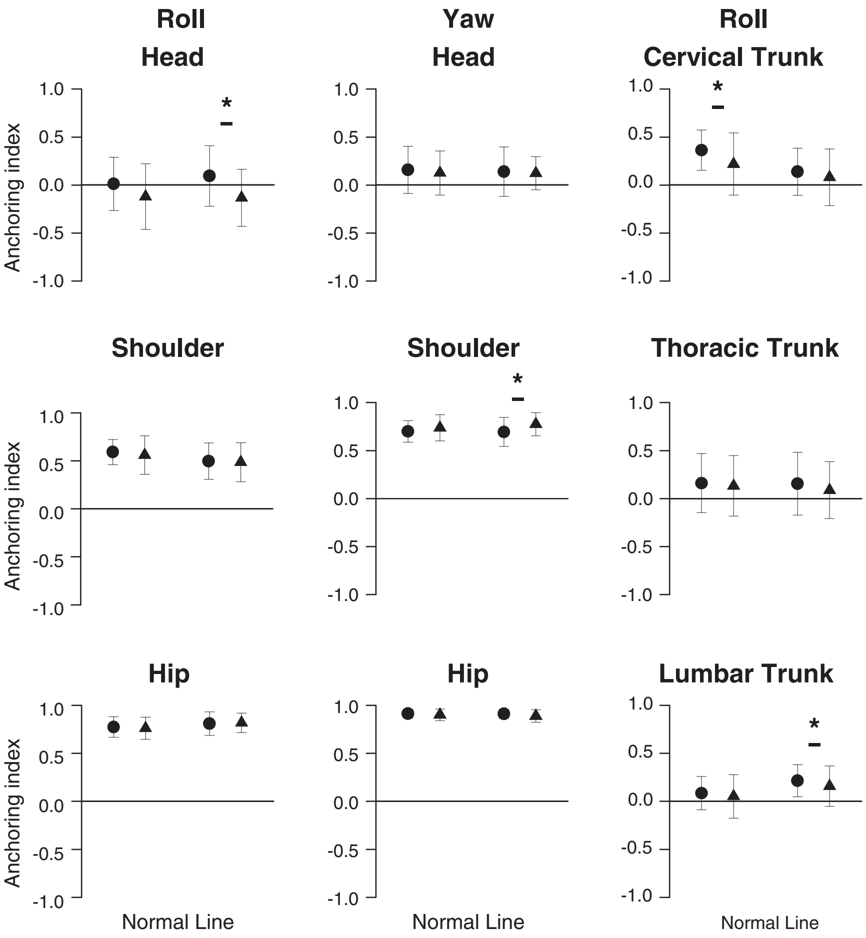

Fig. 3. Mean (SD) roll and yaw anchoring index of the various segments in NGJH (circle) and GJH (triangle) children in both walking conditions (normal and line). Asterisks indicate statistically significant differences.

line, here GJH children instead showed decreased AI of the lumbar trunk ($P = 0.044$) (right part of Fig. 3).

3.1.2. Yaw

No significant differences in the yaw AD of the head, shoulders or hip were observed during normal walking, but during walking on a line a greater yaw AD of the hip was observed in GJH children ($P = 0.042$) (middle of Fig. 2). No differences were found in the AI's around the yaw axis during normal walking, but during walking on a line, GJH children showed significantly greater AI at the shoulder level ($P = 0.041$) (middle of Fig. 3).

3.2. Adults

3.2.1. Roll

For adults, there was no difference between the groups with respect to the roll AD of the head neither during normal walking nor on a line, but at the shoulder level a difference was observed in both walking conditions, showing that adults with GJH had significantly larger AD than NGJH (normal walking: $P = 0.001$; walking on a line: $P = 0.015$) (left part of Fig. 4). As shown in the right part of Fig. 4, GJH adults did also show increased roll AD at the level of the thoracic trunk (normal walking: $P = 0.001$; walking on a line: $P = 0.049$) and lumbar trunk (normal walking: $P = 0.003$; walking on a line: $P = 0.004$) in both walking conditions.

Both groups showed independent stabilization of the shoulder and trunk segments regardless of the walking conditions, but the groups differed regarding to the AI of the shoulder (normal walking: $P < 0.001$; walking on a line: $P < 0.001$) and thoracic trunk (normal walking: $P < 0.001$; walking on a line: $P = 0.031$) in both walking conditions (Fig. 5). In each case AI was lower for GJH adults showing decreased independent stabilization of the shoulder and thoracic trunk.

In addition, GJH adults showed lower roll AI of the cervical trunk when walking on a line ($P = 0.024$) and lower roll AI of the hip during normal walking ($P = 0.023$) as shown in the right and left part of Fig. 5 respectively.

3.2.2. Yaw

No group differences were seen in the yaw AD of the head, shoulder and hip (middle of Fig. 4).

Both groups showed head, shoulder and hip stabilization in space regardless of the walking condition, but GJH adults differed from NGJH adults at the shoulder level by significant higher values of yaw AI in both walking conditions (normal walking: $P = 0.010$; walking on a line: $P = 0.020$) (middle of Fig. 5).

4. Discussion

The use of the anchoring index in the present study provided a useful means of quantifying the dynamic balance of body segments during walking, it also provided evidence for the use of different strategies among the groups of subjects regarding the head orientation. Dynamic balance and movement variability could also have been quantified by calculating e.g. *approximate entropy* for a number of segment angles, however, the current experimental design did not enable such analysis.

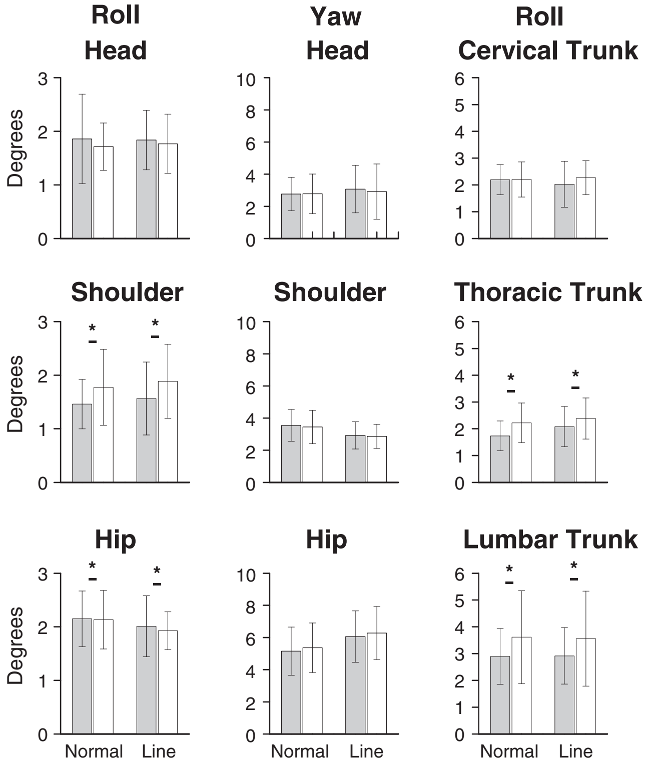

Fig. 4. Mean (SD) of roll and yaw angular dispersions in degrees of the various segments in NGJH (gray) and GJH (white) adults in both walking conditions (normal and line). Asterisks indicate statistically significant differences.

4.1. Children

The present results showed that GJH children walked with different dynamic stabilization strategies and decreased head and trunk stability compared to NGJH children. The roll head AI value in GJH children became negative when the balance was challenged, while it remained positive in NGJH children. This reflected a change, from “articulated” to “en bloc” mode of lateral head stabilization in GJH children when walking on a line. The mode of head stabilization describes the functioning of the neck joints, which make it possible to control the head rotations. The head can be stabilized on the trunk with the neck joints blocked, which is called “en bloc” functioning or the head can be stabilized in space, with the neck joints loose, which is called “articulated” functioning. These concepts can be extended to any couple of consecutive anatomical segments of the body (Assaiante and Amblard, 1995.). The “en bloc” mode of stabilization is associated with decreased attenuation of oscillations between adjacent segments (Mallau et al., 2007), which may explain why decreased lateral head stability was found in GJH children when walking on a line.

One may wonder how it was possible to observe differences between groups in the head and trunk segments while not in the shoulder segment (Fig. 2). An obvious explanation for this could be that the shoulders and the whole shoulder girdle can move in three planes without any concomitant movement of the trunk or head segments (see Fig. 1).

Blocking the head to the trunk during more demanding locomotor conditions concurs with observations in healthy children below the age of seven years and in patients with Parkinson's disease (Assaiante, 1998; Assaiante and Amblard, 1993; Mesure et al., 1999). Assaiante (1998) proposed an ontogenetic model that describes gradual improvement of the neck joint functioning with age during locomotion. Our findings in GJH children match a strategy attributed to children from three to six years of age, who perform “articulated” mode of neck joint functioning only when walking on flat ground but when the level of difficulty increases the strategy changes to “en bloc” functioning. After the age of seven years children become able to perform “articulated” neck joint functioning even when the balance difficulty increases. The latter strategy concurs with the results of NGJH children in the present study so the difference could indicate delayed locomotor development in GJH children compared to NGJH children.

GJH children showed decreased lateral stability of the lumbar and the thoracic trunk in both walking conditions and decreased “articulated” mode of stabilization in the lumbar trunk when walking on a line. The latter difference may explain the decreased lateral stability of the trunk when walking on a line, because decreased “articulated” mode of stabilization leads to less damping of oscillations from the legs. This reduced the stability of the lumbar trunk itself and superior segments.

The decreased lumbar stabilization and the decreased lateral stability of the trunk may have been a consequence of the decreased lateral head stability when walking on a line, because head stabilization in space is important in order to control lateral trunk equilibrium (Assaiante and Amblard, 1993). Decreased lateral stability of the thoracic and lumbar trunk was also found during normal walking. However, this was not accompanied by decreased head stability in the GJH children. Thus, the present results did not exclusively confirm the relationship between decreased head stability and decreased trunk stability. It should though be noted that non-significant decreased lateral stability of the head was found in GJH children during normal walking.

GJH children showed a stiffer (i.e. decreased “articulated” functioning/increased “en bloc” functioning) upper trunk due to the reduced roll AI of the cervical trunk during normal walking. The stiffening could be an adaptation to increased demand of balance during normal walking since previous studies have shown that healthy adults respond to equilibrium difficulty by increasing the “en bloc” functioning of the upper trunk (Mallau et al., 2007; Mesure et al., 1999; Nadeau et al., 2003). A possible mechanism to increase stiffness is to increase the muscle spindle sensibility in general, which has been shown to happen in elderly subjects (van Schaik et al., 1994; Winegard et al., 1997).

4.2. Adults

GJH adults showed decreased lateral stability of the shoulder, lumbar and thoracic trunk in both walking conditions, but it was not associated with decreased stability of the head in any of the walking conditions. The decreased lateral stability of the thoracic trunk may be due to decreased lateral stability of the lumbar trunk but also due to less “articulated” stabilization in the segment itself. Although, both groups showed “articulated” stabilization of every trunk segment regardless of the walking conditions, the GJH adults showed significantly decreased “articulated” mode of stabilization in the thoracic trunk in both walking conditions and responded to the challenging walking condition by decreased articulated mode of the stabilization of the cervical trunk. This response indicated increased demand of balance in GJH adults since previous results have shown that normal adults respond to equilibrium difficulty by decreasing the articulated mode of stabilization in the upper trunk (Mallau et al., 2007; Nadeau et al., 2003). The decreased lateral stability of the shoulder was not associated with decreased stability in inferior segments, but only decreased articulated mode of stabilization in the shoulder itself.

5. Conclusions

Stability of the trunk was decreased in GJH children and adults. It may be a consequence of decreased stability of the head. GJH children showed “en bloc” mode of head stabilization during more demanding locomotor conditions, indicating delayed locomotor development compared to healthy children. GJH adults responded to equilibrium difficulty by showing decreased articulated mode of stabilization in

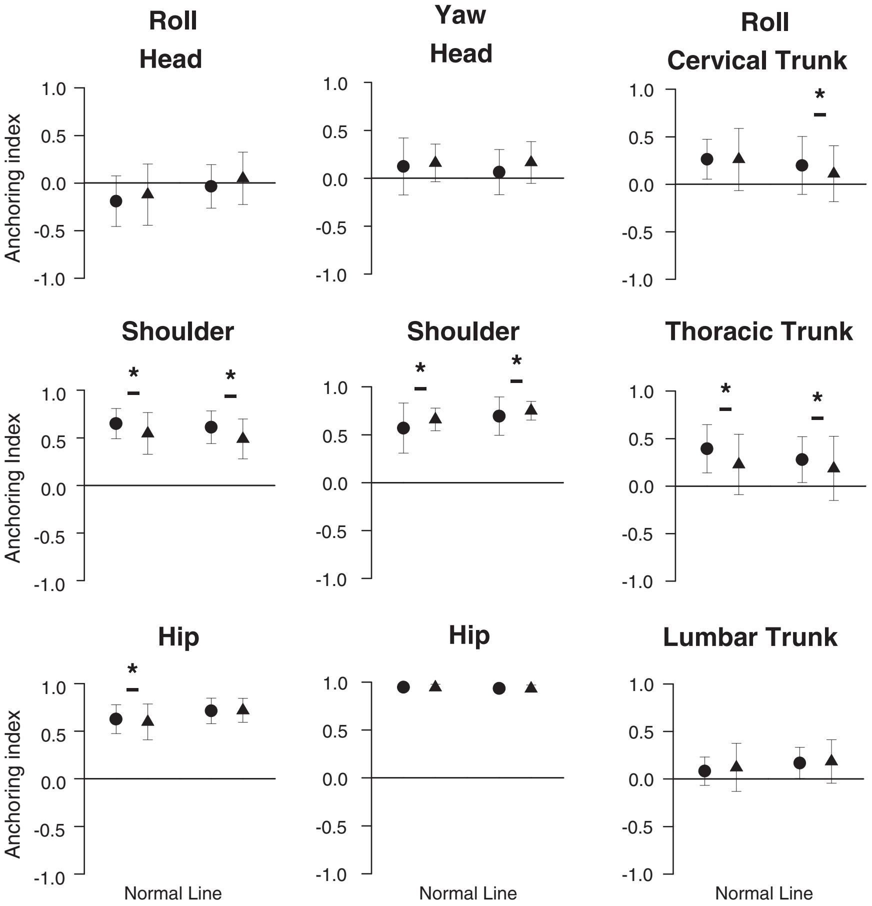

Fig. 5. Mean (SD) roll and yaw anchoring index of the various segments in NGJH (circle) and GJH (triangle) adults in both walking conditions (normal and line). Asterisks indicate statistically significant differences.

the upper trunk, which could indicate increased demand of balance compared to healthy adults. The findings reflect that GJH probably include motor control deficits.

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