Aspects of Isometric Contractions and Static Balance in Women with Symptomatic and Asymptomatic Joint Hypermobility

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Abstract

Objective: The aim of the current study was to identify the differences in strength, balance and muscle activity between women with normal mobility and those with generalized joint hypermobility (GJH) with and without symptoms.

Methods: A total of 195 women, 67 normomobile (NM) and 128 hypermobile (HM), were included in this explorative cross-sectional study, whereby 56 were classified as symptomatically hypermobile (HM-s) and 47 as asymptomatically hypermobile (HM-as). Peak force (Fmax) and rate of force development (RFD) were measured during single-leg maximal voluntary isometric contractions of the knee extensor and flexor muscles in a sitting position. Balance was investigated on a force plate by calculating the anterior-posterior and medio-lateral sway while maintaining a single-leg stance for 15 seconds. During the sway measurements, muscle activity of six leg muscles t-tests, whereas the NM, HM-s and HM-as groups were compared using independent samples ANOVAs with Tukey post-hoc tests (significance level p ≤ 0.05).

Results: While no statistically significant differences were found for Fmax, RFD and postural sway between the three groups, semitendinosus muscle activity showed a difference between the NM and HM (p=0.019) as well as between the NM and HM-as groups (p=0.020).

Conclusions: No clinically meaningful differences were found between the three groups. This might be possible due to the fact that the performance measurements were not sensitive and the motor tasks not challenging enough to detect differences in neuromuscular behavior of the investigated groups.

Keywords: Generalized joint hypermobility; Neuromuscular abilities; Strength; Balance; Questionnaire; Subgrouping; Symptoms

Introduction

Generalized joint hypermobility (GJH) is a condition often seen in the field of rheumatology but mostly underestimated regarding complexity of diagnosis and treatment [1].

GJH depends on age, sex and ethnicity, whereby women are usually more often affected than men and, with increasing age, joint mobility is commonly reduced [2]. In a recent survey conducted by Mulvey et al. [3], the prevalence of GJH in a general population was reported to be 18% with hypermobile subjects having a 40% increased risk of reporting severe chronic widespread pain. Furthermore, recurrent joint dislocation or subluxation, arthralgia, soft tissue injuries and back pain as well as fibromyalgia, chronic fatigue syndrome or early osteoarthritis were associated with the condition [4,5].

The diagnosis of GJH is mainly based on the Beighton Score [6,7]. With the Brighton Criteria [8], which includes additional information on medical history, the Hypermobility Syndrome is identified.

Concerning the diagnosis and treatment of this complex clinical pattern, only little evidence is available, whereby most available publications were based on professional opinions, experience and theoretical knowledge [4,9,10].

The stabilization of a joint is based on both the passive and the active tone. The passive tone includes the elastic properties of the soft tissue, which provide a certain level of stiffness. In several studies, hypermobile subjects showed differences in collagen distribution and reduced stiffness as compared to people with normal mobility [4,11,12]. The active tone is defined by the neuromuscular properties and muscular strength, which has to be split into maximum strength, strength endurance and rate of force development [13].

Activities of daily life such as walking and stair climbing are based on specific strength requirements such as rate of force development [14-16]. In individuals with reduced performance of the musculoskeletal system these activities can be problematic, leading to accidents and often to restrictions in daily life [17].

Multiple authors investigated the musculoskeletal abilities in hypermobile persons and found some differences compared to normomobile persons.
Mebes et al. [18] showed that hypermobile women without symptoms had a higher isometric rate of force development in the knee extensor muscles compared to normomobile women. Another study indicated that muscle strength and functional performance of the lower extremity were reduced in women with the hypermobility type Ehler-Danlos syndrome. Furthermore, these patients demonstrated a significantly lower level of physical activity than controls [19].

In other studies, reduced proprioception and impairment in knee joint position perception was linked to hypermobility [20,21].

A study of Ferrell and colleagues reported reduced balance capability and weaker muscle reflex activity in hypermobile subjects [10]. The same group further reported that an exercise intervention program in hypermobile individuals led to improvements in balance, strength and pain and hence, to an improvement in quality of life [9]. Unfortunately, the exercises of the home-based training protocol were unclear. There is a lack of information on how symptoms and neuromuscular abilities might be linked to each other. Thus, the exact relationship between reduced neuromuscular abilities and symptoms remains unclear.

Therefore the aim of the current study was to identify the differences in strength, balance and muscle activity between women with normal mobility and those with GJH with and without symptoms.

Material and Methodology

Study design

An explorative cross-sectional design was chosen. All measurements were conducted in a single-session, except the questionnaire, which was filled in monthly over a period of six months. Inclusion criteria were checked by an independent physiotherapist. All other investigators were blinded. The study was approved by the local ethics committee and all participants provided written informed consent.

Participants and classification

Recruitment was conducted ad hoc from the staff of the University Hospital Bern, the student body of the Bern University of Applied Science and from additional sources. Prior to the invitation for the laboratory measurements, potential participants were pre-screened by phone. Inclusion criteria consisted of: women aged 18-40 years, body mass index (BMI) ranging from 18-30 kg/m², and no severe pain situation or disability that would restrict the measurements. Exclusion criteria were: surgeries or trauma affecting the lower extremity or disability during stair climbing within six months following the laboratory measurements were classified as asymptomatic. No further classification was possible for twenty-five participants, since they did not return their questionnaires and were therefore excluded from further analyses.

Questionnaire

Symptoms were recorded monthly over a period of 6 months with a face validated questionnaire. On the date of the measurements, the Canadian Occupational Performance Measure Questionnaire (COPM) was completed by each subject [22]. The COPM was developed by occupational therapists as a tool to capture the problems of patients in daily life in a more individual way. In the semi-structured interview, the patient has to identify up to five problematic activities. Then, these activities have to be judged on a scale from 1 to 10 with regard to performance and satisfaction with the performance of each activity.

Based on the declared problem situations two problems were individually defined and additionally three generally known problems in hypermobility were included. These general problems were lifting 10 kg, descending stairs, and remaining in a position longer than 20 minutes. The questionnaire further included a general overview of their impairment, the localization, type, intensity and frequency of the problems of the participants.

Data Collection

Strength: To measure peak force (Fmax) and rate of force development (RFD) during a maximal voluntary isometric contraction (MVIC) of the knee extensor and flexor muscles, participants were seated on a custom-built chair with the knees and hips positioned in 90° of flexion. A sling was attached to the lower end of the tibia (first on the posterior side for the measurement of extension and then on the anterior side for the measurement of flexion) and connected to a one-dimensional strain gauge (KM 1500S; Megatron, Munich, Germany), which was calibrated in Newton (N).

The participants were instructed to push towards extension and flexion, respectively, as fast and as strong as possible and to maintain the force for 5 seconds. Prior to the measurements, participants were allowed to practice the task once. Each measurement was repeated three times with a break of 15 seconds in between.

Force data were sampled at 1 kHz and recorded using the software "ads" (uk-labs, Kempen, Germany).

Balance and muscle activation: To evaluate each individual’s balance, the sway of the center of pressure (CoP) was measured using a force plate (Kistler, Winterthur, Switzerland), while the subjects were standing still on their right leg with eyes open and the knee flexed to an angle of 20 degrees. The subjects were instructed to hold this position as steadily as possible, three times for 15 seconds.

Simultaneously, the activity of the tibialis anterior (TA), medial gastrocnemius (GM), semitendinosus (ST), biceps femoris (BF), vastus medialis (VM) as well as vastus lateralis (VL) muscles was measured using surface electromyography (EMG). Electrode placement and measurement procedure was carried out in accordance with the recommendations of SENIAM [25] and ISEK [26]. In brief, after skin preparation, two pre-gelled AgCl-surface-electrodes (Ambu Blue...
Sensor N, Ambu A/S, Ballerup, Denmark) with a diameter of 5 mm and an interelectrode distance of 2 cm were placed on each of the six muscles. The conductivity criterion was a between-electrode impedance of below 5 kΩ. EMG signals were transmitted via a pre-amplifier (gain: 500; band-pass filter: 10-500 Hz) to a telemetry system (TeleMyo 2400 G2, Noraxon, Scottsdale, Arizona, USA) and sampled in sync with the GRF-data at a frequency of 1 kHz.

**Signal Analysis**

Several signals were analysed using the software “ads” (uk-labs, Kempen, Germany).

**Strength:** Force signals were filtered using a low-pass Butterworth filter with a cut-off frequency of 30 Hz. Peak force (Fmax) and rate of force development (RFD) were extracted from the force-time curves. Fmax was defined as the absolute maximal value within the five seconds of contraction and RFD calculated as the force-difference between 20% and 80% of Fmax divided by the time difference between these two points. The average of the three trials was calculated and normalized to each individual’s body weight in Newton (i.e. Fmax in BW and RFD in BW/s).

**Balance and related muscle activity:** Anterior-posterior (ap) and medio-lateral (ml) sways were calculated and parameterized as follows: mean sway [mm], sway range [mm] and mean sway velocity [mm/s] of the 15 seconds lasting balance test.

Electromyography of all muscles measured was calculated by root-mean-square (15-seconds window) and normalized to the activity during MVC (%MVC). Median frequency (MF: Hz) of each muscles’ activity was taken out of the power spectrum of raw EMG.

All balance and activity variables were averaged over the three trials.

**Statistics**

Statistical calculations were carried out using the software package (SPSS 20, IBMCorp., Armonk, NY, USA).

Descriptive statistics are presented as means and standard deviations for each group. Normal distribution of the variables was confirmed using the Kolmogorov-Smirnov-test.

The differences between the groups NM and HM were evaluated using independent samples t-tests, whereas the differences between the groups NM, HM-s and HM-as were explored using one-way analyses of variance (ANOVs) with Tukey post-hoc tests. P-values ≤ 0.05 were considered statistically significant.

**Results**

The two (NM and HM) as well as the three groups (NM, HM-as and HM-s) were comparable in terms of height, weight and age of the participants (Table 1).

<table>
<thead>
<tr>
<th>Variables</th>
<th>NM (N=67)</th>
<th>HM (N=128)</th>
<th>p-value</th>
<th>NM/HM</th>
<th>NM/HM-s</th>
<th>HM-as</th>
<th>NM/HM-s/HM-as</th>
<th>p-value</th>
<th>p-value</th>
<th>p-value</th>
<th>p-value</th>
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</thead>
<tbody>
<tr>
<td>Age [years]</td>
<td>24.8 (5.4)</td>
<td>25.8 (5.4)</td>
<td>0.234</td>
<td>25.3 (5.4)</td>
<td>25.7 (5.3)</td>
<td>0.701</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Height [cm]</td>
<td>165.7 (5.7)</td>
<td>166.7 (5.9)</td>
<td>0.267</td>
<td>166.9 (6.2)</td>
<td>167.1 (5.4)</td>
<td>0.378</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mass [kg]</td>
<td>60.1 (6.9)</td>
<td>61.2 (6.2)</td>
<td>0.352</td>
<td>60.2 (7.6)</td>
<td>61.6 (7.6)</td>
<td>0.529</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>BMI [kg/m²]</td>
<td>21.9 (2.4)</td>
<td>22.0 (2.7)</td>
<td>0.748</td>
<td>21.6 (2.5)</td>
<td>22.1 (2.5)</td>
<td>0.644</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Right knee flexion [°]</td>
<td>152.1 (5.6)</td>
<td>154.5 (5.3)</td>
<td>0.005</td>
<td>155.6 (4.8)</td>
<td>153.3 (5.5)</td>
<td>0.002</td>
<td>0.001</td>
<td>0.505</td>
<td>0.077</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Right knee extension [°]</td>
<td>4.1 (2.2)</td>
<td>11.6 (2.8)</td>
<td>&gt;0.001</td>
<td>12.2 (3.5)</td>
<td>11.0 (1.8)</td>
<td>&lt;0.001</td>
<td>&lt;0.001</td>
<td>&lt;0.001</td>
<td>&lt;0.001</td>
<td>0.054</td>
<td></td>
</tr>
<tr>
<td>Left knee flexion [°]</td>
<td>149.8 (5.7)</td>
<td>152.4 (6.1)</td>
<td>0.004</td>
<td>153.4 (5.8)</td>
<td>151.6 (6.3)</td>
<td>0.004</td>
<td>0.002</td>
<td>0.22</td>
<td>0.289</td>
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<td></td>
</tr>
<tr>
<td>Left knee extension [°]</td>
<td>4.2 (2.2)</td>
<td>11.5 (2.8)</td>
<td>&lt;0.001</td>
<td>12.0 (3.5)</td>
<td>10.9 (1.7)</td>
<td>&lt;0.001</td>
<td>&lt;0.001</td>
<td>&lt;0.001</td>
<td>&lt;0.001</td>
<td>0.089</td>
<td></td>
</tr>
<tr>
<td>Beighton Score [points]</td>
<td>0.3 (0.5)</td>
<td>7.8 (1.0)</td>
<td>&lt;0.001</td>
<td>7.8 (0.9)</td>
<td>7.7 (1.0)</td>
<td>&lt;0.001</td>
<td>&lt;0.001</td>
<td>&lt;0.001</td>
<td>&lt;0.001</td>
<td>0.74</td>
<td></td>
</tr>
</tbody>
</table>

Table 1: Group characteristics for the normomobile (NM), hypermobile (HM) and symptomatic (HM-s) and asymptomatic hypermobile (HM-as) participants, presented as mean values (standard deviations) and related significance tests (T-Test and One-way ANOVA with Tukey post hoc tests).

**Strength**

Fmax and RFD of the knee extensors and flexors showed neither a significant difference between NM and HM women nor between the three groups (NM, HM-as and HM-s) (Table 2).

**Balance and muscle activation**

No significant differences were found for the measurements of mean sway, sway range and sway velocity between the groups NM and HM or for NM, HM-as and HM-s (Table 3).
Variables | NM | HM | NM/HM | HM-s | HM-as | NM/HM-s/HM-as
--- | --- | --- | --- | --- | --- | ---
Fmax [BW] flexors | 0.33 (0.10) | 0.34 (0.09) | 0.741 | 0.34 (0.10) | 0.34 (0.09) | 0.83
Fmax extensors [BW] | 0.70 (0.14) | 0.71 (0.14) | 0.597 | 0.73 (0.14) | 0.71 (0.12) | 0.406
RFD [BW/s] flexors | 1.15 (0.55) | 1.29 (0.83) | 0.208 | 1.24 (1.11) | 1.32 (0.50) | 0.495
RFD extensors [BW/s] | 3.57 (1.11) | 3.60 (1.18) | 0.849 | 3.50 (1.37) | 3.66 (1.01) | 0.769

Table 2: Peak force (Fmax) and rate of force development (RFD) during a maximal voluntary isometric contraction (MVIC) of the knee extensor and flexor muscles for the groups normomobile (NM), hypermobile (HM) and symptomatic (HM-s) and asymptomatic (HM-as). All variables are normalized to each individual's body weight (BW) and presented as mean values (standard deviations) and related significance tests (T-Test and Oneway ANOVA).

Variables | NM | HM | NM/HM | HM-s | HM-as | NM/HM-s/HM-as
--- | --- | --- | --- | --- | --- | ---
Sway (mean) [mm] ap | 6.0 (1.3) | 5.8 (1.2) | 0.251 | 5.9 (1.1) | 5.8 (1.3) | 0.63
ml | 5.9 (1.2) | 5.8 (0.9) | 0.807 | 5.9 (1.0) | 5.7 (0.9) | 0.504
Sway (range) [mm] ap | 30.3 (6.3) | 29.9 (5.9) | 0.702 | 30.4 (5.6) | 29.6 (6.0) | 0.724
ml | 27.8 (4.7) | 27.8 (3.9) | 0.918 | 28.3 (3.8) | 27.3 (3.9) | 0.502
Sway velocity [mm/s] ap | 35.1 (10.1) | 34.7 (8.4) | 33.8 (7.7) | 34.4 (7.7) | 0.72
ml | 44.2 (10.4) | 44.0 (8.6) | 44.4 (7.8) | 43.5 (8.6) | 0.881

Table 3: Mean sway (mm), sway range (mm) and mean sway velocity (mm/s) in anterior-posterior (ap) and medio-lateral (ml) directions during 15-seconds of single-leg stance for the groups normomobile (NM), hypermobile (HM) and symptomatic (HM-s) and asymptomatic (HM-as) groups, presented as mean values (standard deviations) and related significance tests (T-Test and Oneway ANOVA).

Muscles | NM | HM | NM/HM | HM-s | HM-as | NM/HM-s/HM-as | NM/HM-s/| NM/HM-as
--- | --- | --- | --- | --- | --- | --- | ---
Tibialis anterior | 10.0 (5.7) | 9.7 (4.7) | 0.701 | 9.5 (3.9) | 9.6(4.8) | 0.843
Gastrocnemius medialis | 11.2 (4.4) | 11.5 (4.4) | 0.684 | 11.2 (3.6) | 11.6 (4.9) | 0.586
Semitendinosus | 8.3 (5.9) | 6.3 (4.6) | 0.02 | 5.6 (3.3) | 6.5 (5.3) | 0.019
Biceps femoris | 12.0 (9.6) | 14.3 (4.8) | 0.669 | 10.1 (6.4) | 18.9 (62.6) | 0.425
Vastus medialis | 15.5 (7.7) | 14.8 (7.2) | 0.487 | 14.8 (6.5) | 13.6 (6.2) | 0.291
Vastus lateralis | 15.7 (6.4) | 16.1 (8.2) | 0.741 | 15.5 (6.6) | 16.4 (8.6) | 0.86

Table 4: Activation levels (%MVC) of the six measured lower extremity muscles measured during 15-seconds of single-leg stance for the groups normomobile (NM), hypermobile (HM) and symptomatic (HM-s) and asymptomatic (HM-as), presented as mean values (standard deviations) and related significance tests (T-Test and Oneway ANOVA with Tukey post hoc tests).

In terms of muscle activity, the semitendinosus muscle revealed a significant decrease of mean activation level in the NM compared to the HM group (p=0.020) as well as between the HM-as and NM groups (p=0.019). No further statistically significant difference was found (Tables 4 and 5).
Three different groups were compared in terms of strength measurements: normomobile (NM), hypermobile (HM), and symptomatic hypermobile (HM-s). The mean activation level of the second group was significantly higher than that of the first group, with a p-value of 0.098. Similarly, the mean activation level of the symptomatic group was significantly higher than that of the normomobile group, with a p-value of 0.098. The mean activation level of the symptomatic group was significantly higher than that of the hypermobile group, with a p-value of 0.098. The mean activation level of the hypermobile group was significantly higher than that of the normomobile group, with a p-value of 0.098.

In terms of muscle activity, the semitendinosus muscle revealed a significant decrease of the mean activation level. But in general, the EMG values were very low compared to the activation during maximal muscle contraction and showed a high variance. In all groups, the mean activation level of the BF muscle was higher than the one of the ST muscle. The decrease could be observed between the NM and HM-as groups. A possible explanation for the higher mean activation levels of the BF muscles in the NM group could be that the presence of symptoms led to the development of protective strategies.

The measurement of balance may not be stressful enough for these relatively healthy women, who were not actually seeking medical advice. Thus, participants were asymptomatic at the time of inclusion and able to complete the various tests in the test setting. Therefore, this may represent a sample of hypermobile women who are better able to manage their symptoms than others. However, several of these women had symptoms during the six months following the measurements, meaning that pain and disability may not be constant but rather a sort of on-off phenomenon in this population.

Retrospectively, there were some unclear instructions as how to fill in the questionnaire, especially concerning all the linked questions, and this led to missing data. However, to our knowledge, no validated questionnaire for symptoms like pain and disability in patients with hypermobility in German has been published.

The classification of the three groups based on this questionnaire is disputable. The questionnaire should be revised and validated to be applied to hypermobile subjects. In other studies it was possible to build clusters in the heterogeneity of hypermobility [8]. These two studies the criteria to establish the subgroups were based more on

<table>
<thead>
<tr>
<th>Muscles</th>
<th>NM</th>
<th>HM</th>
<th>NM/HM</th>
<th>HM-s</th>
<th>HM-as</th>
<th>NM/HM-s/HM-as</th>
</tr>
</thead>
<tbody>
<tr>
<td>Tibialis anterior</td>
<td>67.0 (7.7)</td>
<td>65.5 (7.5)</td>
<td>0.195</td>
<td>64.3 (6.9)</td>
<td>66.5 (7.7)</td>
<td>0.161</td>
</tr>
<tr>
<td>Gastrocnemius medialis</td>
<td>63.2 (6.8)</td>
<td>64.1 (6.2)</td>
<td>0.388</td>
<td>63.9 (5.7)</td>
<td>63.2 (6.3)</td>
<td>0.842</td>
</tr>
<tr>
<td>Semitendinosus</td>
<td>64.9 (9.4)</td>
<td>62.6 (6.9)</td>
<td>0.098</td>
<td>63.4 (8.4)</td>
<td>61.6 (9.5)</td>
<td>0.154</td>
</tr>
<tr>
<td>Biceps femoris</td>
<td>41.8 (25.2)</td>
<td>37.3 (25.3)</td>
<td>0.231</td>
<td>36.4 (26.6)</td>
<td>35.9 (25.7)</td>
<td>0.37</td>
</tr>
<tr>
<td>Vastus medialis</td>
<td>53.8 (5.3)</td>
<td>53.3 (5.8)</td>
<td>0.567</td>
<td>52.9 (5.5)</td>
<td>53.7 (5.1)</td>
<td>0.674</td>
</tr>
<tr>
<td>Vastus lateralis</td>
<td>55.9 (6.4)</td>
<td>54.3 (8.5)</td>
<td>0.162</td>
<td>54.1 (6.5)</td>
<td>54.1 (10.8)</td>
<td>0.356</td>
</tr>
</tbody>
</table>

Table 5: Median frequency of the electromyographic signal spectrum (Hz) of the six lower extremity muscles measured during 15-seconds of single-leg stance for the groups normomobile (NM), hypermobile (HM) and symptomatic (HM-s) and asymptomatic (HM-as), presented as mean values (standard deviations) and related significance tests (T-Test and Oneway ANOVA).
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References

