Original Research

The Effect of Joint Hypermobility Syndrome on DOMS and Recovery Time

Nicole F Ostuni[1a], Charles A Marinello[1], Taras Luzhnyy[1], Alexa Pawlikowski[1], Connor Vlasaty[1], Georgios Thomatos[1], Peter C Douris[1]

[1] Physical Therapy, New York Institute of Technology

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Background

Previous research has reported that people with Joint Hypermobility Syndrome (JHS) and Ehlers-Danlos Syndrome (EDS) generally experience a high rate of muscular injury and pain. However, there is limited research comparing the recovery times and length of Delayed Onset Muscle Soreness (DOMS) in individuals with JHS to non-hypermobile individuals in response to exercise.

Hypotheses/Purpose

The purpose of this study was to investigate JHS and its effects on DOMS and its recovery time.

Study Design

Quasi-experimental, observational comparison

Methods

Two groups including a hypermobile group (score >4 on Beighton Scale) and a non-hypermobile group all took part in five-second long standing eccentric bicep curls based using their one- repetition maximum (1-RM) of their dominant arm to failure in order to induce DOMS. Visual analog pain scale (VAS), McGill pain scale, resting arm angle, girth, and the pressure pain threshold, all domains of DOMS, were measured over a five-day period. Results were analyzed using ANOVA with time as the repeated factor.

Results

Both groups experienced DOMS following the eccentric exercise. However, VAS reporting was significantly greater in the hypermobile group compared to the non-hypermobile group and there was a significant difference over time. However, other variables did not reveal any other significant findings between groups.

Conclusion

Individuals with JHS may experience greater DOMS and require more time to recover between treatment sessions. Therapists need to be aware that patients with hypermobility may experience higher pain levels related to exercise, and they need to adjust treatment parameters appropriately.

Level of Evidence

2b

a Corresponding Author:

Nicole Ostuni, PT, DPT, CSCS
Department of Physical Therapy, School of Health Professions, New York Institute of Technology, Old Westbury, New York
Email: nicolefostuni@gmail.com

INTRODUCTION

Joint Hypermobility Syndrome (JHS) is defined as a condition in which an individual exhibits relatively increased or an abnormally large range of motion around a joint and/or joints.[1] JHS is quantified by patients having a score of ≥ 4 on the Beighton Score.[1,2] The Beighton Score has been shown to be an accurate measure for confirming hypermobility in patients.[2] Each point in the Beighton Score represents one of nine possible joints that can present with hypermobility. Research has found that individuals with hypermobility disorders, in particular Ehlers-Danlos Syndrome (EDS), have defective and/or abnormal healing,[3] with a high rate of fatigue.[4,5] Previous authors have reported that people with JHS and EDS experience high rates of muscular injury and pain due to exercise.[6] However, there is limited research comparing the recovery times and length of delayed onset muscle soreness (DOMS) symptoms in individuals with JHS to individuals without JHS in response to exercise. DOMS is defined as muscle pain and soreness that occurs post-exercise which may peak and last for several days after the exercise.[7] It has been documented that the prevalence of JHS was 12.5% overall in college-aged students, 16.2% for women and 8.7% for men.[8]

JHS has been associated with many debilitating features including increased risk for injury, centralized fatigue, centralized pain, inhibition of surrounding muscles, and decreased strength/performance when compared to non-hypermobile individuals.[1,6] Lee et al.[7] investigated the laxity of the anterior cruciate ligament (ACL) and muscle soreness post exercise comparing men and women. They stated that the women reported greater DOMS and the women's ACLs had significantly greater elasticity following exercise when compared to the men. They concluded that women may require more time for musculoskeletal recovery in response to heavy exercise.[7] To et al.[9] studied central vs. peripheral fatigue in participants with JHS and participants without JHS. The participants received electrical stimulation to their biceps to invoke muscle fatigue. Both central and peripheral fatigue was measured in all participants. The control group did not experience central or peripheral fatigue; however the experimental group (JHS) experienced centralized fatigue. This study suggests that individuals with JHS may experience fatigue differently. It has been reported that the transverse abdominis muscle can be inhibited by the surrounding hypermobile vertebrae.[10] The study concluded that decreased muscle activity and increased muscle inhibition exists in muscles surrounding hypermobile joints.[10] Di Stefano et al.[11] investigated on how central sensitization could be the underlying mechanism causing pain in people with JHS and EDS. They stated that pain is a common symptom in patients with the connective tissue disorder, JHS, but the mechanism of why and how are unclear in previous studies. Their study included patients with JHS/EDS that participated in a detailed clinical exam to investigate the somatosensory nervous systems and central sensitization in this population. The authors discovered there was no damage to the somatosensory nervous system of these participants. However, they concluded that most of the subjects experienced widespread pain due to persistent nociceptive input due to joint abnormalities which probably triggers central sensitization in the dorsal horn neurons. Alternatively, Igharno et al.[12] compared the small fiber peripheral nerve fibers and autonomic nervous system of hypermobile EDS subjects to healthy controls. They confirmed that small nerve neuropathy and autonomic nervous dysfunction is a common feature of hypermobile EDS as an underlying pathomechanism of joint pain and dysfunction.

DOMS refers to heightened muscle soreness and pain that peaks and lasts several days after exercise.[7] The intensity of DOMS increases within the first 24 hours post exercise, peaks between 24 to 72 hours, and subsides and eventually disappears in 5 to 7 days.[13] Although resistance exercise has been proven to be an effective intervention for reducing pain and joint instability in hypermobility,[14] individuals with JHS may respond differently to resistance exercise as compared to individuals with normal range of motion due to differences in their nervous system, joint morphology, muscular and ligamentous attachments. The purpose of this study was to investigate JHS and its effect on DOMS and recovery time. The authors' hypothesized that individuals with JHS would experience increased DOMS when compared to non-hypermobile individuals in response to exercise. The results of this study may assist and add to the body of knowledge regarding treating and exercising patients with JHS, as special considerations can be important when treating this population. Patients with JHS may experience greater DOMS and require more time to recover between treatment sessions.

METHODS

PARTICIPANTS

Participants had to be physically active and participate in recreational exercise. Participants were recruited from the greater university community. Participants were selected according to the inclusion/exclusion criteria. The inclusion criteria included: Age range: 18-35 years; Good overall health; Regularly participate in recreational exercise. The exclusion criteria included: Individuals with Ehlers-Danlos Syndrome; Any major musculoskeletal injuries in the prior six months; Any recent traumas that could have led to acute hypermobility or instabilities; Any known disorders that impede recovery/healing time (i.e. Lupus, Rheumatoid Arthritis, Scleroderma); Current elbow pain; Any other health issues that would risk the safety of the subject. The study was approved by New York Institute of Technology Institutional Review Board (BHS # 1636) and the study was registered at www.ClinicalTrail.gov (NCT04934267). The participants were informed of the methods, procedures, risks and were asked to sign the approved consent form prior to starting the study.

PROTOCOL

The research design was a quasi-experimental, observational comparison. Group 1 was the non-hypermobile group and Group 2 was the hypermobile group. The inde-

pendent variables were the two groups and the dependent variables were girth, resting arm angle (RANG), Pain 1-10 Visual Analogue Scale (VAS), Short Form McGill Pain Questionnaire (SF-MPQ 2), and pressure pain threshold (PPT) using an algometer. This battery of measures were utilized to quantify the domains of DOMS because there does not exist a single outcome measure to define it and has been previously utilized in the literarture.[15] All participants took part in an exercise session with eccentric bicep curls based on their 1 repetition maximum (1-RM), which was the highest amount of weight that they could lift concentrically once, using their dominant arm. Both groups performed one set of standing eccentric bicep curls based on their 1RM to failure in order to induce DOMS.[15] Each rep included a timed five second long eccentric component with a metronome and without a concentric component, as the research conductors lifted the weight up concentrically for the participant. The exercise stopped when the participant could not volitionally keep up with the five second count lowering the weight or their form was disrupted. This procedure was previously utilized by Douris et al.[15] Participants were asked to refrain from self-treatment of their DOMS by taking pain medication, using massage or other pain or edema reducing modalities, as well as refraining from any upper extremity exercises during the duration of the study. Prior to exercise, baseline measurements were taken for girth, pressure pain threshold, and RANG. Starting day 2, all measures (including girth of dominant brachium, PPT, VAS, McGill Pain Questionnaire, and RANG) were taken every day at the same time of day, for the following four days. VAS and the McGill Pain Questionnaire were not taken because the subjects were without pain prior to starting the protocol according to the exclusion criteria. Each dependent variable and change in measurements from baseline or day 2 to day 5 post exercise were recorded and used for subsequent data analysis.

MEASUREMENT

Girth: Girth was the measurement of the circumference of a limb (in this study, the midline of the dominant brachium) in centimeters, using a standard tape measure, measuring swelling and/or edema, which commonly occurs during DOMS.[15] A standard tape measure was utilized.

RANG: Resting Arm Angle (RANG) is a ROM measurement in degrees, using a standard goniometer, of the dominant arm (elbow joint) while resting.[15] A standard goniometer was utilized.

VAS Numeric Pain Distress Scale: The 0-10 point VAS Numeric Pain Distress Scale is a commonly employed self-completed scale for the assessment of pain in adults. The scale is marked with 10 equal intervals starting with 0 and ending in 10, to quantify the level of self reported pain. The anchor (0/10) is marked "no pain", the middle (5/10) is marked "moderate pain", and the end (10/10) is marked as "unbearable pain". Greater distances are associated with increased pain levels. In the absence of a gold-standard for pain assessment, the VAS has demonstrated high correlation with a 5-point verbal descriptive scale and a numeric

rating scale for pain (r = 0.71-0.78 and r = 0.62-0.91, respectively.[16]

SF-MPQ 2: The SF-MPQ 2 is a shorter version of the original McGill Pain Questionnaire. It is a multidimensional measure of pain, consisting of two subscales, one for sensory change and one for affective change. The SF-MPQ 2 has been shown to be valid when compared to VAS (r = 0.926) with strong test-retest reliability (ICC = 0.941) when assessing adult populations.[16,17] The SF-MPQ 2 score is the sum of the 22 pain descriptors.

Algometry: Algometry is the assessment of load-dependent tenderness at a specific anatomical site. Tissue tenderness has been conventionally measured by subjecting myofascial structures to external pressure. Algometers serve to quantify pressure pain thresholds and have been shown to be valid up to 80N (r = .990) when compared to force plate output.[18] Additionally, interrater and intrarater reliability were 0.92 (r = 0.87-0.95) and 0.84 (r = 0.73-0.90), respectively.[17] Research suggests algometry may be useful in the assessment of treatment efficacy (14)18. The Force Ten™ FDX Pressure Algometer (Wagner Instruments, Greenwich, CT 06836) was utilized. The procedure for algometric measurement was adapted from Park et al[17] and was standardized according to the following: continuous ascending pressure was at a constant rate, to the midline of the anterior brachium, in order to quantify the individuals pain pressure threshold, measuring in kilogram-force (kgf).[18-20] Pressure was increased at a rate approximately 1 kg/cm^2/s. The participant indicated increased pain by saying the word "ouch," the test was then concluded, and the measurement recorded. The procedure was repeated for a total of three trials; average scores were calculated and recorded.

STATISTICAL ANALYSIS

All values are reported as mean ± standard deviation. Statistical analyses were performed utilizing SPSS for Windows (version 27.0, Armonk, NY).The independent variables consisted of two groups, an experimental group (hypermobile individuals), and a control group (individuals that were not hypermobile with normal ranges of motion). The dependent variables were girth, resting arm angle (RANG), Visual Analogue Scale (VAS), McGill Pain Questionnaire, and pressure pain threshold using an algometer. In order to test the hypothesis, a mixed design ANOVA was performed for each dependent variable with time as the repeated factor. The two main effects were the two groups and time and the interaction effect was the interaction of time and group. The assumption of sphericity was tested using Mauchly's test. In the event that sphericity was violated, a Greenhouse-Geisser correction factor was applied. A priori sample size calculation revealed that 10 subjects were required in each group in order to detect observed differences at a power of 80%. A p value≤.05 was accepted for statistically significant differences.

Table 1. Subject Characteristics (n=24)

AGE (yrs)	25.7 ± 4.0
HEIGHT(cm)	169.4± 10.4
WEIGHT(kg)	68.0 ±13.0

RESULTS

This study included 24 men and women participants. Group1 included 12 subjects, 10 men and 2 women with normal ranges of motion (score of <4 on Beighton Scale). Group 2 included 12 subjects, 10 women and 2 men, with increased ranges of motion (score of ≥ 4 on Beighton Scale and hypermobile in the elbow. The subject characteristics of the twenty four participants are presented in Table 1

The means and standard deviations for all dependent variables are reported in Table 2.

There were significant main effects for time for VAS, McGill, RANG, and PPT with no interaction effects. Both groups responded similarly, with significant changes over the five- day period (main effect of time) which reflects the response to DOMS. The main effect of group was also significant for the dependent variable of VAS (p=0.015) and girth (p=0.037). However, post hoc analysis with independent t-tests between the groups revealed that only the VAS scores were significantly greater in the hypermobile group compared to the non-hypermobile group on Day 2 (p=0.005) with a large effect size (d=1.30) and Day 4 (p=0.037), also with a large effect size (d=0.93). The results of the mixed design ANOVA for all dependent variables are presented in Table 3.

DISCUSSION

The results of the study demonstrated that individuals with JHS may experience and complain of more pain than individuals without JHS as a result of unaccustomed eccentric exercise. It appears from this data that both groups experienced DOMS over the five day period following the eccentric exercise. The study revealed no evidence of significant differences in the level of DOMS between the two groups except for the VAS scores. The McGill scores, RANG measurements, and PPT measurements were all significantly changed over time for all subjects, which is the expected response to unaccustomed or eccentric exercise, and appears to be unrelated to the level of hypermobility of the participants. The results of this study may be explained by Distefano et al.[11] who demonstrated increased perceived pain in hypermobile participants when compared to non-hypermobile participant, as the hypermobile group experienced increased perception of pain as compared to the non-hypermobile group (VAS scores), with no other dependent variables showing a significant difference between the groups. Girth was the only variable that was not significantly changed over time; however, was significantly different between groups, with a higher girth for the non-hypermobile group on average compared to the hypermobile group. The authors of this study are attributing this to a potential gender effect. The hypermobile group included more women and the non-hypermobile group included more men. The current results compare favorably to Lee et al.[7] after inducing muscle soreness through intense squatting, women reported significantly greater VAS scores compared to men. The study also found that women have greater elasticity in their knee joint and should have more time for musculoskeletal recovery. Although they reported differences between the men and the women, they did not take into account of possibility of increased hypermobility prior to exercise in the knee joint in the women as compared to the men which can be inferred by their reporting greater elasticity of the women's anterior cruciate ligament of the knee joint after exercise.

To et al.[9] reported that subjects with JHS experienced more centralized fatigue when compared to subjects without JHS after inducing muscle fatigue to their biceps. However, they did not experience more peripheral fatigue. This central fatigue may contribute or be related to the increased perceived pain that the subjects with hypermobility experienced in this study. Terry et al.[6] concluded that peo-

Table 2. Variables Days 1-5

Variable	Group	Day 1	Day 2	Day 3	Day 4	Day 5
VAS (0-10)	1		2.3±1.5	3.6±1.8	1.9±1.8	0.3±0.9
	2		4.7±2.2	4.7±1.8	3.9±2.5	1.4±1.8
McGill	1		72.3±76.1	86.7±109.9	68.6±118.2	18.2±46.9
	2		54.2±40.8	86.4±65.1	62.9±75.9	19.3±36.6
PPT (kg)	1	2.7±0.9	1.8±0.8	1.4±0.7	1.7±0.8	2.0±0.8
	2	2.6±0.8	1.5±0.8	1.0±0.8	1.4±1.0	1.9±1.1
RANG (deg)	1	23.8±5.7	30.4±7.9	33.0±13.5	31.0±11.8	25.8±6.2
	2	22.9±3.8	35.8±5.5	36.2±7.5	36.4±12.8	28.2±4.6
Girth (cm)	1	31.0±4.5	31.6±4.6	32.2±4.5	31.6±4.5	31.5±4.6
	2	27.7±3.4	28.6±3.4	28.7±3.6	26.3±8.4	28.8±3.5

Values are mean ± standard deviation. VAS- Visual Analog Scale for Pain, McGill- Short Form McGill Pain Questionnaire, PPT- Pain Pressure Threshold, RANG Resting Arm Angle. Group 1- non-hypermobile group, Group 2-hypermobile group

Table 3. Results of Mixed design ANOVA

Variable	Factor	F	p value
VAS	Time	$F_{3,66} = 33.87$	**.001**
	Group	$F_{1,22} = 6.89$	**.015**
	Interaction	$F_{3,66} = 1.84$	.148
McGill	Time	$F_{3,66} = 8.58$	**.001**
	Group	$F_{1,22} = .047$	.830
	Interaction	$F_{3,66} = .200$	.659
PPT	Time	$F_{4,88} = 29.77$	**.001**
	Group	$F_{1,22} = .649$	.429
	Interaction	$F_{4,88} = .452$	.771
RANG	Time	$F_{4,88} = 13.812$	**.001**
	Group	$F_{1,22} = 1.44$	.243
	Interaction	$F_{4,88} = 9.78$	.424
Girth	Time	$F_{3,66} = 33.87$	**.001**
	Group	$F_{1,22} = 4.91$	**.037**
	Interaction	$F_{3,66} = 1.84$	.148

Bold font denotes a significant difference (p<0.05)

ple living with JHS report higher levels of pain, fatigue, and repeated episodes of injury. Their study also indicated that people with JHS report and experience greater anxiety about getting injured or trying to "fit in" and that acceptance by medical professionals help with their long term recovery.[6] Thus, there may be a psychological component to the higher perceived pain levels in the JHS population as well.

There were several limitations of the current study. As mentioned previously, a gender effect was at play, as the non-hypermobile group in the current study mostly consisted of men and the hypermobile group consisted primarily of women. This was especially evident with girth measurements, as the men, on average, had larger arm girths than the women. As previously discussed, there are physiological differences between females and males that affect recovery and pain perception as well.[7] Future studies may attempt to study men or women separately to control for the confounding effect of gender. Some participants reported they were not fully recovered by the end of the five days, and therefore the time limit of five days for assessment in the current study served may be a limitation. Additionally, the authors of this study believe the SF-MPQ 2 may not have been the best outcome measure to include as it incorporates many different pain types that may have not been the most appropriate for this particular study. This study was heavily reliant on many subjective outcome measures as is true of most studies on DOMS. More expensive and objective outcome measures such as ultrasound imaging and blood markers of muscle damage may be warranted.

CONCLUSION

The results of this study indicate that individuals with JHS may experience greater DOMS and require more time to recover between treatment sessions. Therapists need to be aware that hypermobile patients may experience higher pain levels, and so may need to adjust treatment parameters appropriately. This also means that therapists need to listen to their hypermobile patients' feedback carefully regarding their pain levels when prescribing interventions. Resistance exercise has been proven to be an effective intervention for reducing pain and joint instability in hypermobility, but therapists need to monitor and progress it individually for hypermobile patients.

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