Generalized Joint Hypermobility and Lower Extremity Musculoskeletal Biomechanics in Female Athletes

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GENERALIZED JOINT HYPERMOBILITY AND LOWER EXTREMITY MUSCULOSKELETAL BIOMECHANICS IN FEMALE ATHLETES

by

Christopher Geiser, M.S., L.A.T., P.T., A.T.,C.

A Dissertation submitted to the Faculty of the Graduate School, Marquette University, in Partial Fulfillment of the Requirements for the Degree of Doctor of Philosophy

Milwaukee, Wisconsin

May 2023
ABSTRACT
GENERALIZED JOINT HYPERMOBILITY AND LOWER EXTREMITY MUSCULOSKELETAL BIOMECHANICS IN FEMALE ATHLETES

Christopher Geiser, M.S., L.A.T., P.T., A.T.C.
Marquette University, 2023

Generalized Joint Hypermobility (GJH), in lay terms being “double-jointed”, affects 5 to 43 percent of the general population. In severe forms, GJH impacts systems across the body, with cardiovascular, ocular, and musculoskeletal effects that can be quite debilitating. Most of the literature examining GJH is in this severely impacted group of people. However less severe forms of GJH are present in the athlete population in the same proportions, and athletes with GJH are more likely to be injured while participating in activities. They also experience greater time-loss injuries while participating in athletic activities. The movement biomechanics of those with severe forms of GJH have been studied, but studies investigating athletes and athletic-like movements are sparse with varying results. Therefore, the purpose of this dissertation was to investigate the movement characteristics of those with GJH and identify patterns of movement that may put this group of athletes at risk for injury.

Seven collegiate Division 1 women’s Lacrosse athletes from the same team were identified with GJH and evaluated during athletic-like task compared to control participants from the same collegiate team. In the first study, participants performed a maximal countermovement jump while force data were collected to evaluate performance variables between groups. The second study involved both groups of participants performing a bilateral drop jump and a more demanding single-leg land and cut task while kinematic and kinetic data were collected. Finally, the third study used the land and cut task data to create a musculoskeletal model to evaluate ACL strain during the task in each group.

The primary findings of this dissertation were that female division 1 Lacrosse athletes with GJH 1) demonstrated equal performance characteristics with their non-hypermobile teammates, 2) demonstrated similar biomechanics during athletic-like tasks until the task became more challenging on their dominant leg, where they adopted a pattern of less hip and knee flexion and a greater plantarflexor moment, and 3) the pattern of motion adopted during the strenuous land and cut task was not largely an attempt to minimize ACL strain.
ACKNOWLEDGMENTS

Christopher Geiser, M.S., L.A.T., P.T., A.T.,C.

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A special thanks to Dr. Paula Papanek, who inspired me to make the transition to academia and who convinced me that my clinical expertise was worth sharing with students and the field of Athletic Training. Your support both professionally and personally is one of the most important factors in my personal life journey.

I’m also grateful to my participants from the Marquette Women’s Lacrosse team for their time and efforts in obtaining this information, and to Marquette strength coach Maggie Smith, who was my sounding board, provided clinical insight into the problem and practical needs of this population, and inspiration to pursue an academic answer to a very practical problem we were experiencing.

Thanks to my friends and family who supported me through the process, helping with encouragement, support, stress relieving companionship during long runs, and friendly jabs to keep me progressing forward in the process. You kept me sane and gave me outlets for the restlessness that builds up while reading and writing.

Lastly, thank you to my wife and partner in all things, Mary Beth Geiser. You make this all worth it and I love you.
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CHAPTER 1: INTRODUCTION

Generalized Joint Hypermobility (GJH) is a form of joint laxity that affects individuals systemically (Carter & Wilkinson, 1964) and likely occurs from differences in their collagen structure (Russek, 1999). Severe forms of hypermobility are a component of Ehlers-Danlos and Marfan syndrome, which are associated with recurrent joint dislocations and instability at multiple joints. These are generally debilitating conditions with associated disabling musculoskeletal effects, cardiac and vascular issues, and ocular disorders. These individuals are impacted in most aspects of their lives by their hypermobility (Castori & Colombi, 2015; Ross & Grahame, 2011).

Unlike these severe forms of hypermobility, there are individuals with asymptomatic GJH who are not noticeably impacted by this joint laxity during activities of daily living and are often simply referred to as “double-jointed.” A greater incidence of benign GJH is found in females and children, with the overall incidence decreasing with age (Russek, 1999). Therefore, it is not surprising that individuals with asymptomatic GJH are also present in the young female athletic population where 5% to 43% of the female athletes can be categorized as having GJH (Decoster et al., 1999; Konopinski et al., 2012; Pacey et al., 2010). The increased flexibility associated with GJH can be advantageous in certain activities such as ballet, figure skating, or gymnastics. However, a growing body of evidence indicates that while athletes with GJH may not be impacted by their GJH during activities of daily living, they are at greater risk of non-contact knee injury during physical activity, and when injured, athletes with GJH are out for greater amounts of time (Konopinski et al., 2012; Pacey et al., 2010). Hence, the severity of injuries and time lost to injury is greater in the GJH.
group of individuals. A study of 859 West Point Cadets over a 4-year period identified GJH as a one of 4 variables in a predictive model for ACL injury in women (Uhorchak et al., 2003). Furthermore, the consensus statement released following the 2015 ACL Research Retreat VII lists GJH and knee joint laxity as a risk factor for ACL injury and associates this laxity with high-risk landing mechanics (Shultz, Sandra J., Schmitz, Benjaminse, Collins, Ford, & Kulas, 2015).

To understand the differences in why this group of individuals is more frequently and more severely injured during participation in athletics, and to have a chance to develop strategies to decrease the risk of injury, investigators first need to identify factors that predispose this population to injury. Arguably, a major emphasis should be placed on investigating neuromuscular and biomechanical risk factors, as movement-related patterns increase injury risk (Koźlenia & Domaradzki, 2021), but appear modifiable through training interventions (Griffin, L. Y. et al., 2000). Some investigations have identified small differences in how individuals with GJH move compared to controls. In one study, individuals with GJH were noted to have higher muscle activation levels of the vastus medialis, lateralis, and medial gastrocnemius during normal-speed walking, presumably in an attempt to better stabilize a more-lax joint (Schmid et al., 2013). In another study, frontal-plane knee and hip abductor moments were reportedly greater in individuals with GJH during gait (Simonsen, E. B. et al., 2012). However, both of these findings came while evaluating individuals from the general population and not from the athletic population, and evaluated normal-speed gait, which is not a physically demanding task like those encountered in athletics.
Few studies have examined the biomechanics of people with GJH during athletic or strenuous activities. Two studies have examined biomechanical differences between individuals with GJH and otherwise healthy controls during a more athletic-like task. In one of these studies, individuals with GJH absorbed a greater amount of energy at the knee joint compared to the control group during a drop jump landing task (Shultz, S. J., Schmitz, Nguyen, & Levine, 2010). Absorbing more energy at the knee during landing is accomplished by greater use of the quadriceps muscle, which acts antagonistically to the ACL with respect to tibial translation. This finding may help explain the increased risk of ACL injury reported in this population. Still, the subjects with GJH in that study consisted of individuals from the general college population, did not include exclusively high-level athletes, and was limited to a drop vertical jump task, which is not a truly demanding task for athletes. Another study examined a cutting task in a group of individuals who self-reported current participation in cutting or jumping athletic tasks (Hanzlíková et al., 2021). In this study, differences were reported in the kinematics of the dominant leg at the knee, but in a direction that would appear less problematic for individuals with GJH – they demonstrated a smaller knee valgus angle at the knee. However, the subjects were not reported to be high level or elite athletes. None of the previous studies of GJH have included strenuous single-leg tasks, which are particularly more demanding on the hip joint (Geiser et al., 2010) and may affect males and females differently (Weinhandl et al., 2015). Moreover, most previous studies of GJH have not specifically excluded individuals in the GJH group who were experiencing joint pain (Galli et al., 2011) (Simonsen, Erik B., Tegner, Alkjær, Larsen, Kristensen, Jensen, Remvig, & Juul-Kristensen, 2012). Pain impacts musculoskeletal function in many ways
(Hodges, 2011), and while it is relevant to GJH to study individuals with symptomatic laxity, such as Ehlers-Danlos Syndrome, the presence of pain or symptomatic laxity cloud the results of these studies and confound conclusions about the effect of GJH on movement variables that contribute to injury risk in a young, healthy athletic population. Individuals with GJH can and do participate in high level athletics without symptoms, and while their increased risk of injury has been demonstrated (Konopinski et al., 2012; Pacey et al., 2010), the impact of GJH on their movement characteristics has not been reported in a representative group of subjects.

The difference in injury rates and knee injury risk in females with GJH is likely influence by many factors. One factor could be differences in physical performance capabilities between those with GJH and controls. GJH likely occurs from genetic-based differences in collagen synthesis and quality (Castori et al., 2017). These differences have an effect at the joint level, resulting in laxity at the joint, which is how these individuals are classified into the GJH group. However, there are also differences at the muscle fiber level that affect muscle contractile performance (Ottenheijm et al., 2012) in single muscle fiber studies. These differences in muscle at the cellular level may influence the athletic performance at the whole-body level, and may account for differences in injury rate and injury risk through either a lack of sufficient muscular support for the joints, or even a mismatch in level of competition. **Specific Aim 1** defines whether there are performance differences in elite Division 1 female lacrosse athletes with GJH versus controls from the same team as they perform the foundational athletic task of jumping. The ground reaction force and impulse were examined during a countermovement jump
to determine if GJH results in performance differences between groups during this maximal task.

Understanding the neuromuscular and biomechanical control strategies, especially in relation to the task dependency of those strategies during athletic movements in individuals with GJH improves assessment of the risk of injury during pre-activity screenings and allows for the development of specific conditioning activities that target potentially deleterious movement patterns. **Specific Aim 2** foundationally defines movement patterns and biomechanical differences between individuals with GJH and controls in an elite female athletic population. Efforts during submaximal and more demanding athletic-like lab-based tasks were evaluated for group differences in lower extremity biomechanics: Hip, knee and ankle angles and moments in three planes. Statistical parametric mapping (SPM) was utilized to examine the entire ground contact phase of these activities (Friston et al., 1994), rather than discreet variables which only evaluate one out of many data points and may not compare similar time points across groups.

GJH is a known risk factor in ACL injury in women (Uhorchak et al., 2003). Further, a consensus statement identifies GJH as a risk factor for ACL injury and associates this laxity with high-risk landing mechanics (Shultz, Sandra J. et al., 2015). Monitoring the ACL directly during athletic tasks is currently not possible. The ligament can be visualized on MRI, but current MRI technology is very limited for activities involving any motion during collection. This makes it impossible to collect the ballistic and aggressive tasks needed to challenge elite level athletes. Computer simulations can be used to build upon the motions collected in the lab and evaluate the length and
position of the ACL during these tasks. For specific aim 3, the biomechanical data collected during the dominant leg single leg cut task in aim 2 was used to create a musculoskeletal model. Using that model, the strain of the ACL was calculated during the land and cut task and compared across groups. **Specific Aim 3** identifies whether the landing mechanics collected during the land and cut task creates different ACL strains between the groups, therefore impacting the ACL injury risk. It also provides evidence of whether the strain of the ACL might be a factor influencing the motor control system of those with GJH when performing these strenuous tasks.

Thus, when viewed as a series, these experiments answer whether there are differences in the way individuals with GJH move during strenuous activity, and whether those differences result simply from physical performance abilities or are an attempt to control the strain on the anterior cruciate ligament during activity. The answers to these aims can guide how injury preventive measures in individuals with GJH can be directed in the future to minimize injury, and to appropriately advise individuals with GJH about the mechanical issues they face participating in high level activities.
CHAPTER 2: JUMP PERFORMANCE VARIABLES ARE NOT DIFFERENT IN FEMALE DIVISION 1 LACROSSE ATHLETES WITH GENERALIZED JOINT HYPERMOBILITY

ABSTRACT

Generalized joint hypermobility (GJH) affects 5 to 43 percent of the population, including a similar proportion of athletes. Individuals with GJH sustain both greater frequency and severity of injuries than the non-GJH population. It is also known that strength deficits may play a role in injury risk, and there is evidence that those with GJH may exhibit strength deficits. However, this has not been examined in high-level athletes. This investigation assessed maximal jump performance in female collegiate Lacrosse athletes from the same team with and without GJH to evaluate for performance and strength differences. Time series data and calculated discreet variables from ground reaction force were compared, demonstrating no strength or performance differences between the groups. Among females competing athletically at the same level, it does not appear that there are strength deficits contributing to the increased lower extremity injury risk in those with GJH.

INTRODUCTION

Generalized joint hypermobility (GJH) is defined as a form of systemic joint laxity (Carter & Wilkinson, 1964) thought to occur from genetic difference in the collagen makeup of these individuals (Russek, 1999). Severe forms of hypermobility are a component of Ehlers-Danlos syndrome and Marfan syndrome. Unlike these often-debilitating forms of hypermobility, individuals with GJH are generally not impacted during activities of daily living and are often referred to in common vernacular as
“double-jointed.” A greater incidence of GJH is found in females and children (Russek, 1999). GJH is also reflected in the athletic population, where the incidence of GJH among adolescent female athletes is estimated to be as high as 43% (Konopinski et al., 2012; Pacey et al., 2010).

There is growing evidence that athletes with GJH are at greater risk of knee injury during athletic participation (Pacey et al., 2010). Overall, they are injured more frequently and miss participation for longer periods of time when injured (Konopinski et al., 2012). Differences in muscle contractile function have been noted in people with severe forms of hypermobility when compared to controls (Voermans et al., 2007). Individuals with Ehlers-Danlos Syndrome Hypermobility Type were found to have lower quadriceps peak force on isometric testing. Weakness at the hip and knee have been noted to increase the risk of ACL injuries (Crowell et al., 2021; Khayambashi et al., 2016; King et al., 2021; Zebis et al., 2022). If athletes with GJH also demonstrate strength deficits as a group, that weakness may account for some of the differences in injury risk for those with GJH, as lower strength values and weakness have been equated with injury risk in women (Augustsson & Ageberg, 2017). However, the individuals with GJH noted to have muscle weakness by Voermans et al were quite impacted by their GJH, noted pain during testing, and were not athletic or even active individuals. The decreased activity level or the pain during testing alone could account for differences in muscle performance. Investigations to determine if a component of the increased injury risk noted in GJH could be attributable to differences in physical performance have not been done in athletes, and those conducted on non-athletic individuals have reported inconsistent findings with other confounding factors, such as joint pain, limited mobility
or lower activity level (Mebes et al., 2008). Therefore, the purpose of this investigation was to evaluate for differences in physical performance in NCAA DI female Lacrosse athletes between GJH and matched controls. While it limits participant numbers and study power, individual subjects from a single collegiate team were recruited to minimize variability between groups that are typically difficult to control for, such as time of year, weather, training routines, competition timing and level, conditioning schedule and coaching techniques. The null hypothesis was that there were no differences between the two groups.

There are many methods for assessing performance in athletes. In recent years it has become relatively common to monitor performance, physical readiness and recovery in athletes by means of serial countermovement jump testing. A maximal countermovement jump (CMJ) utilizes muscular strength, proprioception, and the coordination of multiple muscle groups in a relatively simple but relevant task to most athletic endeavors. Multiple variables can be pulled from CMJ data to assess components of muscular performance (Heishman et al., 2020; Hori et al., 2009). These variables have the sensitivity to reliably identify differences within athletes (Claudino et al., 2017; Kipp et al., 2016). More specifically, maximal strength measures were correlated to CMJ ground reaction forces (GRF) and center of mass (COM) velocity patterns in female collegiate lacrosse athletes (Haischer et al., 2022). A CMJ therefore provides an expedient, consistent, and relevant method for evaluating differences in physical performance.
METHODS

After acquisition of informed consent, all healthy uninjured women’s lacrosse players on a NCAA DI University roster were tested for GJH according to the number of positive signs on the Beighton and Horan Joint Mobility Index (Beighton & Horan, 1969; Juul-Kristensen, B., Rogind, Jensen, & Remvig, 2007), which is an evaluation of 9 specific movements including hyperextension of the knees and elbows past 10 degrees, dorsal flexion of the 5th digit past 90 degrees, thumb and forearm approximation, and the ability to touch the palms flat to the floor with the knees in full extension (Figure 2.1). Previous studies used either 4 or 5 positive signs for inclusion in the GJH group (Pacey et al., 2010; Simonsen, Erik B. et al., 2012; Smith et al., 2012). For this investigation, we used the higher value of five or more positive signs for inclusion in the hypermobile group to account for small day-to-day variations across the menstrual cycle in the number of positive signs (Shultz, S. J. et al., 2012). After a warmup period, participants performed three trials of a CMJ standing with each leg on a separate AMTI force plate, with the instructions to keep hands on their hips and jump “as high as they can.” One practice jump was required prior to data collection, more were allowed if the participant wanted them to feel comfortable. Force data were collected at 960 Hz in Vicon Nexus software and filtered with 4th order Butterworth filter with a cutoff frequency of 12 Hz in a custom Matlab program. Right and left leg vertical reaction forces were summed and the following dependent variables were calculated and exported from Matlab with custom written software for statistical analysis: Vertical Jump Height, modified Reactive Strength Index, Peak Vertical Force, Time to Peak Force, Peak Concentric Force, Peak Eccentric Force, Peak Rate-of-Force Development, Peak Rate-of-Force Development
Figure 2.1 Beighton and Horan Joint Mobility Index (BHJMI). The 9 clinical signs comprising the assessment of the BHJMI.

normalized to Body Weight, Peak Concentric Rate-of-Force Development, Peak Eccentric Rate-of-Force Development, Take-off Velocity, Peak Power, Force at Peak Power, Velocity at Peak Power, Positive Work, Negative Work, Eccentric Time and Concentric Time (Hori et al., 2009; Kipp et al., 2016). Group comparisons were made between the three trial averages for each individual (Claudino et al., 2017). Independent t-tests were used to compare all means between groups with significance set at \( p < 0.05 \).

As any significant differences in variables would reject the null hypothesis, no correction for multiple comparisons was made in an effort to minimize Type II error possibility. Many of the discreet CMJ variables may correlate with each other, such as Peak Take-off
Velocity and Jump Height, but all were included for the exhaustive evaluation of these data.

RESULTS

Thirty-three players were screened with the BHJMI for GJH. The experimental (GJH) group consisted of 9 players (19.4±1.0yrs, 71.0±9.4kg, 165.1±7.1cm) with 5 or more Beighton’s signs (range 5-7, mean 6.0). Seventeen controls (CTRL: 19.9±1.2yrs, 65.3±7.0kg, 167.6±3.0cm) from the same team were included with zero or one Beighton’s sign (range 0-1, mean 0.1). Data for all variables met the assumptions of normality and equal variance for parametric testing. No differences were found between GJH group or controls for anthropometric data.

No differences were noted between groups for any of the CMJ GRF curves in the SPM analysis (Figures 2.2, 2.3). No differences were detected in the analysis of discrete CMJ variables (Table 2.1).
Figure 2.2 Composite averages of the GRF (xBodyWeight) during countermovement jumps for GJH (Red) and CTRL (Black).

Figure 2.3 SPM Analysis of GJH vs CTRL GRF
Table 2.1 Statistical analysis of discreet variables calculated from CMJ GRF.

<table>
<thead>
<tr>
<th>Measure</th>
<th>GJH mean (Stdev)</th>
<th>CTRL mean (Stdev)</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Body Mass (kg)</td>
<td>71.7 (7.5)</td>
<td>67.2 (5.8)</td>
<td>0.11</td>
</tr>
<tr>
<td>Jump Height (m)</td>
<td>0.22 (0.03)</td>
<td>0.23 (0.05)</td>
<td>0.47</td>
</tr>
<tr>
<td>Modified Reactive Strength Index</td>
<td>0.23 (0.04)</td>
<td>0.25 (0.07)</td>
<td>0.34</td>
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<tr>
<td>Peak Force (N/kg)</td>
<td>20.5 (0.9)</td>
<td>21.0 (2.0)</td>
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<tr>
<td>Time to Peak Force (s)</td>
<td>0.74 (0.15)</td>
<td>0.77 (0.15)</td>
<td>0.56</td>
</tr>
<tr>
<td>Peak Concentric Force (N/kg)</td>
<td>20.5 (0.9)</td>
<td>21.0 (2.0)</td>
<td>0.47</td>
</tr>
<tr>
<td>Peak Eccentric Force (N/kg)</td>
<td>18.6 (1.7)</td>
<td>18.7 (2.0)</td>
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</tr>
<tr>
<td>Peak Rate of Force Development (N/sec)</td>
<td>5163 (851)</td>
<td>6067 (2961)</td>
<td>0.38</td>
</tr>
<tr>
<td>Normalized Peak Rate of Force Development (N/kg/sec)</td>
<td>72.5 (12.8)</td>
<td>88.7 (40.9)</td>
<td>0.26</td>
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<tr>
<td>Peak Concentric Rate of Force Development (N/kg/sec)</td>
<td>3534 (1515)</td>
<td>3339 (1464)</td>
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<tr>
<td>Peak Eccentric Rate of Force Development (N/kg/sec)</td>
<td>4811 (895)</td>
<td>5723 (2921)</td>
<td>0.37</td>
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<tr>
<td>Take-off Velocity (m/sec)</td>
<td>1.95 (0.21)</td>
<td>2.04 (0.25)</td>
<td>0.37</td>
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<tr>
<td>Peak Power (Watts/kg)</td>
<td>36.5 (3.8)</td>
<td>39.1 (6.2)</td>
<td>0.28</td>
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<tr>
<td>Force at Peak Power (N/kg)</td>
<td>18.6 (1.4)</td>
<td>19.2 (1.4)</td>
<td>0.29</td>
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<tr>
<td>Velocity at Peak Power (m/sec)</td>
<td>1.97 (0.18)</td>
<td>2.02 (0.21)</td>
<td>0.51</td>
</tr>
<tr>
<td>Positive Work (Joules/kg)</td>
<td>5.84 (1.19)</td>
<td>5.79 (0.92)</td>
<td>0.90</td>
</tr>
<tr>
<td>Negative Work (Joules/kg)</td>
<td>1.98 (0.65)</td>
<td>1.62 (0.51)</td>
<td>0.13</td>
</tr>
<tr>
<td>Eccentric Time (sec)</td>
<td>0.22 (0.04)</td>
<td>0.21 (0.05)</td>
<td>0.78</td>
</tr>
<tr>
<td>Concentric Time (sec)</td>
<td>0.32 (0.05)</td>
<td>0.29 (0.04)</td>
<td>0.11</td>
</tr>
<tr>
<td>Eccentric:Total Time (ratio)</td>
<td>0.41 (0.05)</td>
<td>0.41 (0.06)</td>
<td>0.82</td>
</tr>
<tr>
<td>Center of Mass Flexion Range-of-motion (m)</td>
<td>0.31 (0.08)</td>
<td>0.27 (0.07)</td>
<td>0.23</td>
</tr>
<tr>
<td>Eccentric Range-of-motion (m)</td>
<td>0.15 (0.04)</td>
<td>0.12 (0.04)</td>
<td>0.12</td>
</tr>
<tr>
<td>Concentric Range-of-motion (m)</td>
<td>0.40 (0.09)</td>
<td>0.37 (0.05)</td>
<td>0.37</td>
</tr>
</tbody>
</table>

DISCUSSION

This exhaustive evaluation of jump performance variables in individuals with GJH failed to identify any differences between those participants with and without GJH. This differs from the work done on isolated muscle fibers in hypermobile individuals where lower peak torque was reported in isolated muscle fibers, but greater twitch torque...
in individuals with a more severe form of hypermobility versus controls (Voermans et al., 2007). Another single fiber study found that peak force was equal between groups, but that resting tension in the muscle fibers was greater in hypermobile individuals (Ottenheijm et al., 2012). Greater resting tension is thought to be the reason that rate-of-force development was higher in GJH groups (Mebes et al., 2008) as well as a higher single-fiber twitch torque (Voermans et al., 2007).

Despite the differences noted at the single fiber level in individuals with hypermobility (Ottenheijm et al., 2012; Voermans et al., 2007), the current study did not detect any performance differences between those with GJH and controls during a large, functional movement. While single muscle fiber performance and whole-body activity are obviously different tasks, the discrepancy in findings may also be due to the cognitive preparation for motor activity in this study. The CMJ task allows for coordinated muscle activity between large groups of muscles. This may allow differences in muscle performance at the single-fiber level to be compensated for by timing, coordination, or some other factor during the jump. Perhaps future studies of performance variables in GJH should consider a “reactive” component to capture any differences in muscle resting tension or twitch response that a planned CMJ may not have effectively evaluated.

This study also conflicts with the literature examining those with Ehler’s Danlos hypermobility type and quadriceps strength (Fatoye et al., 2009; Voermans et al., 2007), where those with hypermobility were found to have lower quadriceps strength. However, the individuals tested in these studies were functioning at a fairly low level compared to the controls and were symptomatic from the extent of their connective tissue disorder. Juul-Kristensen reported equal strength in children with GJH (10-year-olds), but some
decrease in quad strength in adults with GJH (Juul-Kristensen, Birgit et al., 2012).

Mebes reported equal strength in those with hypermobility, but a greater rate of force development in the hypermobile group (Mebes et al., 2008). In this study we found no differences in any of the performance variables. However, our GJH subjects were free from injury and functioning at a collegiate division 1 level in Lacrosse. The only discerning difference between the subjects and the controls was their score on the BHJMI. Most other sources of variability were controlled for by including only members from the same team – time of year, training / competition schedules, training philosophy, climate, and competition level. It is likely that strength deficits noted between hypermobile individuals and controls in previous studies were impacted by factors other than GJH.

The current results examined individuals from the same competitive team. The athletes in this study all play Division 1 Lacrosse, all perform at the same level, and were selected to the team because of their ability to perform at this level. They are therefore different than subjects evaluated in previous studies. This in itself is interesting, demonstrating that at least some individuals with GJH are capable of jump performance at the same level as their non-GJH counterparts. Thus, by demonstrating that these athletes with GJH can perform a CMJ equal to non-GJH individuals, this study allows us to consider other variables that may predispose GJH individuals to greater injury risk.

CONCLUSIONS

Participants with GJH participating at a high level in athletics do not appear to exhibit differences during isolated measures of athletic performance when compared to similar level teammates. Thus, the reported differences in injury rates and severity of
injury in people with GJH appears to be driven by factors other than those related to
countermovement jump performance and the muscular characteristics needed to perform
CMJ’s. Future studies must examine other factors that could drive the risk of lower
ex tremity injury in individuals with GJH so that scientifically based effective
interventions can be designed.
CHAPTER 3: GENERALIZED JOINT HYPERMOBILITY IS ASSOCIATED WITH TASK-DEPENDENT INCREASES OF AT-RISK KNEE MECHANICS IN FEMALE ATHLETES

ABSTRACT

Individuals with generalised joint hypermobility (GJH) are at greater risk for non-contact knee injury. This study evaluated the landing biomechanics of female athletes with GJH for differences their non-GJH teammates. All athletes from a Division 1 university women’s Lacrosse team were screened for GJH using the Beighton and Horan Joint Mobility Index (BHJMI). The GJH group was composed of seven athletes with a BHJMI score of five or more; the control group was seven athletes with scores of zero or one. Double- and single-leg landing mechanics were collected with a motion analysis system and two force plates. Variables were extracted for the hip, knee, and ankle, and analyzed using Statistical Parametric Mapping for between-group differences.

While no differences were found for the Drop Jump task, in the more strenuous single leg CUT task, females with GJH demonstrated less knee flexion, lower internal knee abductor moment, and a greater ankle plantar flexor moment on the right/dominant leg. The pattern of less knee flexion and greater plantar flexor moment has been previously associated with non-contact anterior cruciate injury and may partly explain the greater risk of knee injury in athletes with GJH. This is particularly interesting as these findings were specific to the more strenuous single-leg task.

INTRODUCTION

Generalised Joint Hypermobility (GJH) is a form of joint laxity that affects individuals systemically (Carter & Wilkinson, 1964) which may occur from differences in their collagen structure (Russek, 1999). A growing body of evidence indicates that
while athletes with GJH may not be impacted by their GJH during activities of daily living, they are at greater risk of non-contact knee injury during physical activity (Konopinski et al., 2012; Pacey et al., 2010). For example, a study of 859 West Point Cadets over a 4-year period identified GJH as a one of 4 variables in a predictive model for ACL injury in women (Uhorchak et al., 2003). Furthermore, consensus statements list GJH and knee joint laxity as risk factors for ACL injury and associates this laxity with high-risk landing mechanics (Shultz, Sandra J. et al., 2015). Moreover, the injuries sustained by athletes with GJH tend to be more serious in nature than individuals without GJH (Konopinski et al., 2012; Pacey et al., 2010).

To understand the differences and decrease the risk of injury, investigators first need to identify the mechanics that predispose this population to knee injuries. Arguably, a major emphasis should be placed on investigating biomechanical risk factors as movement-related patterns increase injury risk but appear modifiable through training interventions (Griffin, K. M. et al., 2000). However, few studies have examined the biomechanics of people with GJH during athletic or strenuous activities. In one study, individuals with GJH were noted to have higher muscle activation levels of the vastus medialis, lateralis, and medial gastrocnemius during normal-speed walking, presumably in an attempt to stabilize a lax joint (Galli et al., 2011). In another study, frontal-plane knee and hip abductor moments were reportedly greater in individuals with GJH during gait (Simonsen, Erik B. et al., 2012). However, both findings came while evaluating individuals from the general population and not from the athletic population, and evaluated normal-speed gait, which is not a physically demanding task like those encountered in athletics.
Other studies have examined biomechanical differences between individuals with GJH and otherwise healthy controls during a drop jump task, but the subjects in these studies were not highly trained athletes. In one of these studies, individuals with GJH absorbed a greater amount of energy at the knee joint compared to the control group (Shultz et al. 2010). Absorbing more energy at the knee during landing is accomplished by greater use of the quadriceps muscle, which acts antagonistically to the ACL with respect to tibial translation. Still, the subjects with GJH in that study consisted of individuals from the general college population, did not include exclusively high-level athletes, and was limited to a drop vertical jump task. Previous studies of GJH have not included strenuous single-leg tasks, which are particularly more demanding on the hip joint (Geiser et al., 2010) and may affect males and females differently (Weinhandl et al., 2015). Understanding the biomechanical strategies, especially in relation to the task dependency of those strategies during athletic movements in individuals with GJH would improve assessments of the risk of injury during pre-activity screenings and allow for the development of specific conditioning activities that target potentially deleterious movement patterns. These interventions could then be specifically targeted to the population in need, thereby making efficient use of valuable training time and resources.

The purpose of this investigation, therefore, was to elucidate differences in movement patterns by evaluating joint kinematics and kinetics during both a single and double leg activity in athletic individuals with GJH. Specifically, this investigation examined the hypothesis that women collegiate level lacrosse athletes with GJH would exhibit different lower extremity joint mechanics during the landing phases of a drop jump and single-leg land and cut task than their non-GJH counterparts from the same
team, with the null hypothesis that there would be no differences between groups for any variable.

METHODS

After acquisition of institutionally approved written informed consent, all healthy, asymptomatic, and currently uninjured female lacrosse players on the roster of an NCAA Division I University team were tested for GJH utilizing the Beighton and Horan Joint Mobility Index (BHJMI) (Beighton & Horan, 1969). The BHJMI is a clinically convenient method for assessing GJH that requires no expensive equipment and has been used for classifying individuals with GJH in many previous investigations, with acceptable reliability and validity (Boyle et al., 2003; Juul-Kristensen, B. et al., 2007). The GJH group consisted of players scoring five or greater on the BHJMI. Inclusion into the control group (CTRL) required a score of one or zero. Players with a BHJMI score of two through four were excluded from the study. Subjects were also excluded if they had a history of significant lower extremity injury resulting in more than a month of time out of athletic participation, or if they were withheld from play due to a trunk or lower extremity injury in the last 3 months.

During the biomechanical testing session participants were fitted with 18 individual 9.5mm reflective markers and 3 marker clusters (Figure 3.2). The individual markers were attached to the ASIS and PSIS, greater trochanters, medial and lateral knees, medial and lateral malleoli, and the first and fifth metatarsal heads. The marker clusters attached to the thigh, shank, and foot segments consisted of four, four, and three 9.5mm markers, respectively. After the collection of a static trial, all individual markers, except the ASIS and PSIS markers were removed for the remainder of the data collection
and segments were tracked via the marker clusters. Participants were then asked to perform two tasks in a counterbalanced order: 1) A single leg land and cut (CUT) on each lower extremity, and 2) a double-leg drop jump (DJ). The CUT task (Figure 3.1) involved standing on a box away from the force plate, jumping forward and landing on the right leg, then cutting immediately 90 degrees to the left (Geiser et al., 2010). The height of the box was equal to their vertical jump height (Weinhandl et al., 2015), measured by first performing a maximum countermovement jump and tracking a PSIS marker. The box was set back from the force plate a distance equal to each participant’s maximum single leg stride length, which was measured by having them stand on the center of the force plate and take one stride toward where the box would be positioned. This task has been used and described previously in the evaluation of single leg landing and cutting mechanics (Geiser et al., 2010) and was chosen because of the strenuous nature of the task while still allowing expedient data collection in a controlled setting. To avoid biasing subjects, the same instructions to ‘land and quickly cut to the left’ were given to each participant. Participants were corrected if they rotated and turned to face left instead of performing a side-step to the left. This task was then repeated on the opposite side. Right and Left side cuts were purposefully counterbalanced in their order to minimize any learning effect as the task was completed.

For the DJ task, participants stood on the same height box as the CUT, but with the front edge of the box lined up with the edge of the force plates, such that subjects dropped forward and down, landed with each foot on one of the force plates simultaneously, and finished with a maximal vertical jump. Participants were required to
practice each task at least twice and were allowed to practice more if needed. The same instructions to ‘land and quickly jump as high as they can’ were given to each participant. As these were skilled high-level athletes who have completed these tasks for previous screenings, no participant needed more than 3 repetitions to achieve confidence with either task. Trials were discarded and re-collected if the participant failed to reach the force plate, did not land completely on the force plate, did not achieve a 90-degree cut, or faltered in their attempt to complete the task. Tasks were performed in a purposefully counterbalanced order. A minimum of 3 good trials were collected for each task, with no participant needing more than 8 trials to accomplish the CUT or 4 trials to accomplish the DJ.

![Figure 3.1 The “CUT” task.](image)

The three-dimensional position data of the reflective markers were captured at 120Hz with a 14-camera motion analysis system (Vicon Inc. Oxford, UK). The ground reaction forces (GRF) during the stance phase of both tasks were simultaneously collected with two floor embedded force plates (AMTI Corp. Watertown, MA) that
recorded at 960 Hz. The collected kinematic and kinetic data were both filtered with a 4th order low-pass Butterworth filter that had a cutoff frequency of 12 Hz. A 3D kinematic model was built in Visual 3D software (C-motion Inc., Germantown, MD) based on the following specifications: The hip joint centers were defined as 25 percent of the horizontal distance between trochanter markers, knee joint centers as midpoint of medial and lateral knee markers, and ankle joint centers as midpoint of medial and lateral malleoli markers (Robertson et al., 2004) (Figure 3.2). This model was then used to calculate the joint angle and joint moment data during each trial. Net joint moments (NJM) were calculated using an inverse dynamics approach (Bresler & Frankel, 1950; Kadaba et al., 1990), normalized to the athlete’s body weight (Jones et al., 2014), and are presented as internal NJM. The dependent variables of interest included in the analysis were the hip, knee, and ankle angles and NJM in the sagittal and frontal planes. The data in the transverse plane was not used for analysis given the generally unreliable nature of transverse plane surface marker data. The dependent variables were extracted and analyzed during the ground contact time of each task, identified when the GRF exceeded 30 N through the point where the GRF diminished below 30N.

Data were analyzed using Statistical Parametric Mapping (SPM) over the ground contact phase for both joint angles and net joint moments in each plane for each joint. The significance level was set at $\alpha = 0.05$. Statistical analyses were performed in Matlab using the SPM function. The small number of subjects available on this one team for each group meant the study was powered to detect differences with an effect size Cohen’s d of 1 or greater at a $p<0.05$ level.
RESULTS

A total of 38 female lacrosse athletes were assessed for BHJMI score. Seven players (19 ± 1 yrs, 66.0 ± 6.1 kg, 167.3 ± 3.3 cm) with GJH (BHJMI score mean 6.0, range 5-7) were identified. Seven players (20 ± 1 yrs, 62.1 ± 7.1 kg, 165.3 ± 7.3 cm) with a BHJMI score of 0 or 1 (BHJMI score mean 0.14 ± 0.38) were identified for the control group. No between group differences were noted for any anthropometric data (p<0.05). All participants were Right-leg dominant.

For the Drop Jump in the sagittal plane, left knee flexion moment was greater in the GJH group (p=0.017, α=0.05, t threshold=3.744) from roughly 5 to 20 percent of the ground contact time (Figure 3.4). In the frontal plane on the right (dominant) side only, the GJH group demonstrated a lower knee adduction angle (p=0.046, α=0.05, t threshold=3.555) and a lower internal knee abductor moment (p=0.003, α=0.05, t
threshold=3.722) during roughly the first 20 percent of the ground contact time (Figures 3.7 and 3.8). For the CUT, no differences were noted on the left (non-dominant) leg (Figures 3.5, 3.6, 3.9 and 3.10). For the right (dominant) leg when it was the stance leg for the CUT, differences were detected. In the sagittal plane, the GJH group demonstrated less right hip and knee flexion angles during the CUT task (Figure 3.5) between roughly 25 and 50 percent of the ground contact time (Hip: $p=0.035$, $\alpha=0.05$, $t_{\text{threshold}}=3.097$; Knee: $p<0.001$, $\alpha=0.05$, $t_{\text{threshold}}=3.567$) and a greater right ankle plantar flexor net joint moment (Figure 3.6) between roughly 60-75% of the ground contact time ($p<0.001$, $\alpha=0.05$, $t_{\text{threshold}}=3.578$). In the frontal plane, the GJH group demonstrated a lower internal abduction net joint moment at the knee (Figure 3.10) between roughly 5-15% of the ground contact time ($p=0.009$, $\alpha=0.05$, t-threshold=3.714).
Figure 3.3 Sagittal plane angles during the DJ task. GJH=dashed red, CTRL= solid black, Shaded region is the 95% confidence interval. No statistical differences were present between groups. SPM graphs for each variable can be found in Appendix 2.1.
Figure 3.4 Sagittal plane moments during the DJ task. GJH=dashed red, CTRL= solid black. Shaded region is the 95% confidence interval. Surrounding red box indicates statistical differences between groups within that time-curve variable. SPM graphs for each variable can be found in Appendix 2.2.
Figure 3.5 Sagittal plane angles during the CUT task. GJH=dashed red, CTRL= solid black. Shaded region is the 95% confidence interval. Surrounding red box indicates statistical differences between groups within that time-curve variable. SPM graphs for each variable can be found in Appendix 2.5.
Figure 3.6 Sagittal plane moments during the CUT task. GJH=dashed red, CTRL= solid black. Shaded region is the 95% confidence interval. Surrounding red box indicates statistical differences between groups within that time-curve variable. SPM graphs for each variable can be found in Appendix 2.6.
Figure 3.7 Frontal plane angles during the DJ task. GJH=dashed red, CTRL= solid black. Shaded region is the 95% confidence interval. Surrounding red box indicates statistical differences between groups within that time-curve variable. SPM graphs for each variable can be found in Appendix 2.3.
Figure 3.8 Frontal plane moments during the DJ task. GJH=dashed red, CTRL= solid black, Shaded region is the 95% confidence interval. Surrounding red box indicates statistical differences between groups within that time-curve variable. SPM graphs for each variable can be found in Appendix 2.4.
Figure 3.9 Frontal plane angles during the CUT task. GJH=dashed red, CTRL= solid black, Shaded region is the 95% confidence interval. No statistical differences were noted between groups. SPM graphs for each variable can be found in Appendix 2.7.
**DISCUSSION**

The novel findings from this study highlight task- and group-dependent differences in landing biomechanics between subjects with and without GJH on the
dominant leg. More specifically, this study identified that athletes with GJH use a more upright landing position than controls when performing a CUT task on the dominant leg, but not a DJ task. These differences in the GJH group are consistent with movement biomechanics that have been implicated in the ACL injury mechanism, or that increase ACL injury risk (Boden, B. P., Dean, Feagin, & Garrett, 2000; Hewett et al., 2005; Shimokochi et al., 2013; Thomas et al., 2010). While the subject groups were small, the differences reported here represent large effect sizes to be detected in this size study.

This study examined female athletes sampled from the same athletic team at the same point in their season, which offers a high degree of control for differences in training regimens and other environmental factors which are notoriously difficult to control for in most studies. Collectively, these results indicate that GJH may impact movement mechanics in a way that may increase the risk of significant knee injury in this population, especially during more demanding single-leg tasks.

The GJH group landed more upright during the CUT on their dominant leg, exhibiting less hip and knee flexion during the landing phase of the CUT (Figure 3.5), and a greater plantar flexor moment (Figure 3.6). These differences were not present in the DJ. A more upright landing position has been noted during the analysis of in-game ACL injuries (Boden, B. P. et al., 2000), has been associated with a greater risk of non-contact ACL injury (Hewett et al., 2005; Shimokochi et al., 2013), and has been noted to contribute to the actual ACL injury mechanism during cadaveric testing (Oh et al., 2012). It has also been demonstrated using dual plane fluoroscopy techniques that the ACL strain increases sharply after ground contact during landing and decreases with subsequent knee flexion (Taylor et al., 2011). It is interesting that the differences
observed in the GJH group in this study occurred only during the CUT task, where the knee extensor moments were on average close to double the DJ. The overall musculoskeletal demands were greater during the single-leg than a double-leg task as evidenced by the larger NJM in the knee extensors and ankle plantar flexors across both groups during that task. This finding suggests that the CUT task poses a greater risk of ACL injury than the DJ in this group, and further suggests that the differences in landing mechanics seen in the GJH group during the CUT are driven by the intensity of the task. This is in agreement with Dai et all in which they demonstrated less knee flexion during higher intensity cutting and landing tasks (Dai et al., 2019). Given that a more upright landing posture is consistent with reported mechanisms and risk factors for non-contact ACL injury (Boden, B. P. et al., 2000), these results may help explain why individuals with GJH experience more severe injuries and a greater number of knee injuries during intense athletic tasks. The current results also indicate that studying more demanding tasks may provide better insight into the injury mechanisms and risks in this group of athletic individuals with GJH who are otherwise healthy and functioning at a high level.

Only the dominant leg demonstrated differences during the CUT task. The dominant leg was defined as the leg the athlete would use to “kick a ball for accuracy and distance”. During the kicking motion, the non-dominant leg is the stance leg, which is therefore more accustomed to functioning in the closed chain, planted position. This may explain the differences noted here in side-to-side function. Athletes may therefore be less accustomed to planting and cutting, such as the motion during our CUT task, on the dominant leg. However, this was not the case in our controls, who adapted to the CUT task on both sides. This is not the first study to identify differences between dominant
and non-dominant lower extremities in the GJH population (Hanzlikova et al., 2021), so it is possible that there are differences in function that impact injury risk and should be further examined and possibly addressed with rehab or conditioning activities. Lacrosse would appear to be a fairly symmetric sport with respect to demands on the lower extremity, unlike soccer where there may be a preference for kicking the ball with one leg or the other, differentiating function of the lower extremities during the sport. However, the upper extremity “handedness” when handling the Lacrosse stick may contribute in some way to differences in demands and function of the lower extremities that are detected in this study.

The GJH group exhibited greater plantar flexor NJM than CTRL’s during the CUT on their dominant leg. There were no differences during the DJ task. A task-dependent increase in the magnitude of the plantar flexor moment during the CUT task in the GJH group is an interesting and novel finding that may also be relevant to ACL injury risk. Greater plantar flexor NJM have been previously associated with a more flexed landing posture and have thus been suggested to be a component of a landing strategy that ameliorates the risk of ACL injury (Shimokochi et al., 2013). Moreover, since ankle sagittal or frontal plane motion did not differ between the two tasks in the current study, greater plantar flexor NJM during the CUT likely points to greater work done and energy absorbed at the ankle joint, effectively reducing the magnitude of knee joint loading, which would also alleviate some ACL risk (Boden, Barry P., Torg, Knowles, & Hewett, 2009; Meyer et al., 2008). Conversely, in studies comparing male and female landing mechanics, females have been found to absorb more energy at distal joints during landing, suggesting that this type of landing strategy may contribute to a greater risk of
ACL injury than employing a more proximal (i.e., hip-focused) landing strategy (Decker et al., 2003; Schmitz et al., 2007). Further, landing with a more distal energy absorption strategy has been previously linked to more risky landing mechanics throughout the lower extremity (Norcross et al., 2010). Admittedly, the role of plantar flexor NJM in relation to ACL loading is not well understood as some simulation models suggest that the gastrocnemius can act as an antagonist to the ACL whereas the soleus can act as an agonist to the ACL (Mokhtarzadeh et al., 2013). The exact effects of the task-dependent differences in plantar flexor NJM in relation to ACL injury risk therefore remain cloudy and should be the focus of further investigation, especially in the GJH population.

Individuals with GJH experience less internal knee abductor NJM than controls during the landing phase across tasks. A larger internal knee adductor moment during a DJ is commonly thought of as a risk factor for ACL injury (Hewett et al., 2005) and is a component within the ACL injury mechanism during cadaveric testing (Oh et al., 2012). Decreasing the knee abductor moment on the other hand brings the moment around the knee closer to zero, meaning less stress on the knee. Another possible explanation for the findings in this study is that because of the uncertain nature of the demanding CUT task, athletes with GJH adopted a “safer” landing strategy at the knee, minimizing the frontal plane moment and transferring load to the plantar flexors, essentially performing a “landing” then a “cut” as two specific tasks rather than a “land and cut” task. If this is the case, it is again interesting that this occurred only on the dominant leg and only in the GJH group during this task and thus may represent an attempt to mitigate injury risk by those with GJH.
There are several limitations of the current investigation. First, all subjects were recruited from a single sport team which limits the generalizability of the present results to different athletic teams and environments. The current study should therefore be replicated in a broader more diverse athlete population. However, this is also a strength of the study as it serves to minimize variability due to other external factors, such as training regime, competition schedule, and many other environmental factors that are difficult to control for. Second, the sample size is small, which increases the possibility of both type I and type II errors. Statistical Parametric Mapping helps address this issue and to minimize the type I error possibility by considering the variability across each specific signal and determining an individual t-threshold for each variable. This also means that only large effect sizes can be statistically detected, which increases confidence in the findings. Lastly, BHJMI scores can vary over the course of the menstrual cycle (Shultz, S. J., Levine, Nguyen, Kim, Montgomery, & Perrin, 2010) and the current investigation did not control for the phase of the menstrual cycle during which testing occurred. However, all subjects were screened for hypermobility using the BHJMI and initially placed into groups, which were confirmed a second time on the day of testing. The groups therefore accurately represented the classification into GJH or control group on two different days, including the day of testing. Moreover, BHJMI scores vary by less than 1 positive sign across the cycle (Shultz, S. J. et al., 2010), so the groups would not likely differ an appreciable amount on alternate days. We also used the higher, more stringent of the common cutoff thresholds for inclusion in the GJH group, choosing to use a cutoff of 5 instead of 4 on the BHJMI assessment.
CONCLUSIONS

The results for the current study indicate that high-level female athletes with GJH demonstrate a more upright landing pattern during a CUT task on their dominant leg than during a DJ task when compared to non-GJH controls. In addition, the GJH group landed the CUT task with a greater plantar flexor moment. In combination, these differences are consistent with patterns that increase the risk of knee injury, specifically ACL injuries, and may partially explain the greater rate and severity of injuries reported in individuals with GJH during participation in athletic events, while the physiological reasons for these differences will require further investigations. The intensity of the task appears to drive the difference in mechanics.
CHAPTER 4: ACL STRAIN IS NOT A LARGE FACTOR IN THE CONTROL OF A SINGLE-LEG LAND AND CUT TASK IN FEMALE ATHLETES WITH GENERALIZED JOINT HYPERMOBILITY

ABSTRACT

Individuals with Generalized Joint Hypermobility (GJH) get injured more frequently and more seriously during activity. They demonstrate task-dependent differences in knee mechanics which may drive that injury risk. The reason they exhibit these differences in mechanics is unknown, but may represent an attempt by the individual to minimize ACL strain during demanding tasks. In this study, knee joint kinematics and strain for the Anterior Medial (AMB) and Posterior Lateral (PLB) bundle of the Anterior Cruciate Ligament (ACL) were calculated using a modified OpenSim model for a group of Division I Collegiate female athletes during a land and cut task. Group comparisons were made between those with Generalized Joint Hypermobility (GJH) and non-hypermobile controls (CTRL) using Statistical Parametric Mapping (SPM). The GJH group demonstrated less knee flexion but no difference in the other planes at the knee nor in ACL AMB or PLB strain during parts of the ground contact time. This indicates that those with GJH may adopt a more cautious landing strategy at the knee with less flexion, but it does not appear this strategy is motivated by the underlying sensation of protecting their ACL strain in a more mobile knee joint during demanding tasks.

INTRODUCTION

Individuals are characterized as having Generalized Joint Hypermobility (GJH) when they have a greater than normal range-of-motion (ROM) at multiple joints (Hakim & Grahame, 2003), often described in lay terminology as “double-jointed”. GJH is
represented in the general population at a frequency between 8 and 43% of people. GJH incidence is higher in young individuals and in females, but is not limited to just those demographics. Individuals with GJH who participate in athletics and strenuous physical activity are more at-risk for knee injury (Pacey et al., 2010) as well as more severe injuries per incidence (Konopiski et al., 2012). Previous studies have examined whether those with GJH move differently during functional tasks with inconsistent findings. Alsiri et al. reported a “stiffening” of the movement patterns during gait in those with GJH (Alsiri et al., 2020), possibly in an attempt to control joint play. Similar alterations in joint mechanics were noted during a drop jump landing, with the GJH group demonstrating increased work absorption and stiffness about the knee (Shultz, S. J. et al., 2010). During an unanticipated cutting maneuver however, the pattern of joint stiffening in those with GJH was not observed (Hanzlikova et al., 2021). They reported a small decrease in ankle plantarflexion angle and an increase in knee external rotation angle during this task in those with GJH.

Others have examined the relationship between ACL elongation and knee kinematics (Nagai et al., 2019; Shelburne et al., 2005). These studies suggest that an elongation of the ACL occurs during the early stance phase of close chain activities, with the ACL undergoing approximately 4-16% elongation as compared to its resting length (Beynnon & Fleming, 1998; Nagai et al., 2019; Shelburne et al., 2005). This range was fairly consistent across studies regardless of activity, model, or collection techniques (Nagai et al., 2019). There were no specific studies that modeled the ACL elongation or strain in those with GJH.
The motor system relies on multiple proprioceptive inputs to generate a motor response (Riemann & Lephart, 2002) in the control of motion and response to external challenges. At the knee this includes the ACL, which supplies sensory information to the central nervous system (Riemann & Lephart, 2002) and appears to impact motor activity around the knee (Poul Dyhre-Poulsen & Krogsgaard, 2000). The joint laxity associated with GJH may alter the tension in the ACL at rest and during activity, thus altering the motor responses and control of the knee during dynamic activity (Zhong et al., 2021). Individuals with GJH may alter their movement patterns to minimize the ACL load to protect the knee from anterior translation during strenuous physical activity (Zeng et al., 2022). However, knee mechanics and modelling of ACL length during strenuous activities in those with GJH have not been evaluated.

The goal of this investigation was to examine differences in knee kinematics during a single leg land and cut activity in GJH and controls by means of a musculoskeletal model to determine if the modelled strain of the ACL during this activity were different between groups. This may provide a partial explanation for the goals motivating motor control differences noted in knee kinematics in the GJH group.

METHODS

Motion capture data from a previous investigation were utilized with known statistical differences in a single leg landing task between GJH and Controls. Briefly, in that study all members of a collegiate women’s Lacrosse athletes were screened for GJH using the Beighton and Horan Joint Mobility Index (BHJMI) (Beighton & Horan, 1969) with a cutoff score of 5 or more for inclusion in the GJH group. Control subjects were used with a score of 0 or 1 on the same scale. Subjects performed 3 trials of a land and
cut task (CUT) off a box normalized to maximum vertical jump height utilizing their
dominant kicking leg as the land and cut leg. 3D kinematic and kinetic data were
collected via a 14 camera Vicon system (Vicon Inc. Oxford, UK) at 120 Hz with kinetic
data collected at 960 Hz (AMTI Corp. Watertown, MA). The CUT task involved jumping
forward from a box, landing on the dominant leg, then cutting 90 degrees. The box height
was scaled to match each subject’s maximum countermovement jump height and set a
distance from the force plate equal to their maximum forward stride length as described
in chapter 3. Data were filtered using a 4th order low pass Butterworth filter, cutoff
frequency of 12 Hz. Marker data were tracked in Vicon Nexus to ensure complete marker
data across the entire trial.

For this investigation, the data were then imported into OpenSim 3.3 (Delp et al.,
2007). The static trial was used to scale a generic musculoskeletal model to each
participant’s dimensions, geometry and body mass. The OpenSim model was used with a
modified knee (Xu et al., 2015), which included three rotations and three translations at
the knee and separate antero-medial and postero-lateral bundle origins and insertions for
the components of the ACL (Figure 4.1). Inverse kinematics were used to calculate joint
angles by minimizing the weighted sum of squared differences method between model
and experimental marker data. Ligament elongation data were calculated for each frame
of the cut task utilizing the origin and insertion of the ACL in the open sim model. The
Anterior Medial Bundle (AMB) and the Posterior Lateral Bundle (PLB) were both
normalized to their resting length during the standing trial (Ro=1). Ligament strain were
calculated using the formula “Strain % = ((Frame length – resting length) / resting
length)”. Data were analyzed for group differences using statistical parametric mapping
(SPM) across the time series length data (Friston et al., 1994) to identify periods where the calculated t-value was above the trial-specific critical threshold for differences (p<0.05).

![Knee model from OpenSim (Xu et al., 2015)](image)

**Figure 4.1** Knee model from OpenSim (Xu et al., 2015)

**RESULTS**

A total of 38 female lacrosse athletes were assessed for BHJMI score. Seven players (19 ± 1 yrs, 66.0 ± 6.1 kg, 167.3 ± 3.3 cm) with GJH (BHJMI score mean 6.0, range 5-7) were identified. Seven players (20 ± 1 yrs, 62.1 ± 7.1 kg, 165.3 ± 7.3 cm) with a BHJMI score of 0 or 1 (BHJMI score mean 0.14 ± 0.38) were identified for the control group. No between group differences were noted for any anthropometric data (p<0.05). All participants were right leg dominant.

Knee flexion data from the modified OpenSim model closely matched the output of the previous model built in visual3D (figure 4.2). In both models, GJH demonstrated
less knee flexion (sagittal plane) on their dominant (right) leg during the land and cut task (t=3.480, α=0.05, p<0.001), with the difference occurring roughly between 25-35% of the stance phase (Figure 4.2). While the group differences were noted in knee flexion angle between groups, SPM analysis revealed no differences in ACL strain in either the AMB or the PLB during the ground contact phase (Figure 4.3, 4.4, 4.5, 4.6).

Analysis of the discreet value group averages for peak AMB and PMB strain also revealed no statistical differences between groups. However, the GJH group had lower values for peak AMB strain (GJH=1.150±0.017; CTRL=1.162±0.015), which is an effect size of d=0.80, a difference which this study was not powered to detect statistically.

Figure 4.2 OpenSim (left) vs Visual3D (right) modelled Sagittal plane knee angle data for GJH (red) vs Controls (black), SPM analysis underneath for each.
Figure 4.3 Right ACL AMB strain during the CUT task. GJH=dashed red, CTRL= solid black, Shaded region is the 95% confidence interval.

Figure 4.4 SPM analysis of right ACL AMB strain.
Figure 4.5 Right ACL PLB strain during the CUT task. GJH=dashed red, CTRL= solid black, Shaded region is the 95% confidence interval.

Figure 4.6 SPM analysis of right ACL PLB strain.

**DISCUSSION**

From chapter 3 of this dissertation, as the task became more challenging, those with GJH adopted a more upright landing strategy, namely less knee flexion on their
dominant lower extremity compared to controls without GJH. In this chapter, we investigated whether this upright landing strategy was driven by or related to protective posturing for the ACL in this population. Specifically, did individuals with GJH alter their lower extremity biomechanics to minimize strain of the ACL. We hypothesized that they may be more protective of their joints that have more mobility, or based on previous literature, if the pattern of landing may be a “stiffening” of the lower extremity to protect the ligamentous structures (Alsiri et al., 2020). However, the musculoskeletal model output did not differ for the ACL anterior medial bundle or the posterior lateral bundle strain, indicating that the differences in knee angle were not largely driven by an attempt to limit the ACL strain during controlled movements.

A stiffer landing, one which utilizes less knee flexion during the weight absorption phase, has been demonstrated to be a risk factor for ACL injury (Boden, B. P. et al., 2000; Ford et al., 2005; Oh et al., 2012; Shimokochi et al., 2013) for a number of reasons. Less knee flexion means a greater compressive force is applied to the knee by the GRF, which in itself is a factor in ACL injuries (Boden, Barry P. et al., 2009; Podraza & White, 2010; Vacek et al., 2016). Additionally, in a more knee extended position the contact point on the femoral surface is flatter, resulting in a propensity to slide posteriorly on the tibia when compressed, versus landing on a more flexed knee where the rounder posterior portion of the femoral surface is in contact with the tibia (Boden, Barry P. et al., 2009; Meyer et al., 2008). This is a common sex difference identified in landing and cutting tasks between males and females (Boden, B. P. et al., 2000; Boden, Barry P. et al., 2009; Vacek et al., 2016). In the current study, all participants were female, thus presumably already more at risk of ACL injury. The
participants with GJH demonstrated less knee flexion as a group, even among all female participants. This movement pattern alone makes those with GJH more at risk for ACL injury than the general female Lacrosse athlete population (Hewett et al., 2005; Shimokochi et al., 2013). However, there is no consensus on why females demonstrate this pattern of movement, nor why those with GJH appear to land with an even stiffer knee than those without. Based on the modelled ACL strain in the current study, it does not appear that limiting ACL strain is a large motivating factor for minimizing knee flexion angle during landing in those with GJH.

The model demonstrated a lengthening of the ACL AMB between 15.0 percent for GJH and 16.2 percent for CTRLs during the CUT task across groups. There are no previous studies reporting modelled ACL strain during this type of strenuous task. The ACL strain reported here is greater than the ACL strain reported in previous studies examining gait, which ranged from 6.7% during running (Nagai et al., 2019) to 13% during normal walking gait (Taylor et al., 2013). There are obvious task differences between the studies, which likely accounts for the greater magnitude in the ACL strain values reported in this investigation.

In addition to the task differences, there are differences in the methods from each of these studies. In the current study, we used skin-based reflective markers to generate the kinematic data, whereas the gait studies used biplanar fluoroscopy of the knee, which is inherently more accurate as it is not subject to skin movement artifact. Additionally, Nagai et al normalized the ACL AMB length to the length of the ACL on MRI scan, whereas our model used the ACL AMB and PLB length calculated during the static trial.
as the normalized length. This difference in baseline measure may also result in different dynamic strain values throughout the movement.

Investigations that examine ACL length over isolated knee range of motion (ROM) report the longest values for AMB length near full extension, and don’t show shorter lengths until after 60 degrees of flexion (Foody et al., 2023; Li et al., 2005; Wu et al., 2010). This differs from the findings in the CUT task, as the ACL AMB strain was highest in the mid stance in both GJH and CTRLs, when knee flexion was at a maximum of between 60 and 75 degrees, and shorter lengths / less strain at the beginning and end of the ground contact phase when knee sagittal plane angle was between 0 and 20 degrees (Figure 4.3). In addition to examining different tasks, differences in strain patterns were possibly because of the tri-planar motion at the knee, along with the muscle forces acting at the knee during the CUT task versus isolated knee flexion/extension motions in the isolated range studies.

The number of subjects in this study was small, a result of our desire to minimize external variability by keeping the subjects to one team during a single season and time point. This lowered the power of the study and the ability to detect small group differences. An analysis of discreet values for peak ACL AMB strain reveals that, while not statistically different, average strain values between groups was lower in the GJH group by 1.2% (15.0% increase length over resting in GJH vs 16.2% in Controls), which is an effect size of d=0.80 and could still represent a sizeable effect of GJH on ACL strain during this task. This is a finding that should be considered in future studies to replicate and confirm our findings, as well as powering studies to detect smaller, more subtle differences in ACL strain. We recruited all participants on a single team to control many
external variables that are notoriously difficult to account for. Expanding the number of participants in the current investigation would have increased the study power to identify smaller effect sizes, but this would have been at the expense of controlling many of the external factors surrounding the performance of high-level athletes. It is possible that the variability present across many populations of athletes, training cycles and activities, time of year, geographic location and many other factors would make imperceptible the significant findings in this study.

CONCLUSIONS

Female athletes with GJH responded to a challenging task on their dominant leg by landing in an arguably more cautious landing pattern – more upright and tentative before cutting to the opposite side as compared to controls without GJH. We created a musculoskeletal model of these participants to investigate whether the less-flexed landing position may represent a subconscious attempt to protect the ACL from strain during the task. However, neither the AMB nor PLB of the ACL experienced greater strain than controls during that activity. While landing with less knee flexion is an independent risk factor for ACL injury, we do not yet have a clear answer why participants with GJH landed using this strategy at the knee.
CHAPTER 5: CONCLUSIONS

Individuals with Generalized Joint Hypermobility (GJH) experience injuries, specifically knee injuries, more frequently than the general population. When they do get injured, the injuries are more severe, and they miss out on their activities longer than in individuals without GJH. The literature examining the musculoskeletal function of individuals with GJH to date has focused largely on those who are severely impacted by their joint laxity, or on those who are simply general functioning members of society. There are very few studies examining the impact of GJH on the athletic population, specifically high-level athletics. The purpose of this dissertation was to explore biomechanical underpinnings for the higher knee injury risk specifically in a group of individuals with GJH functioning at a high level in a competitive collegiate Division 1 athletic program in the United States. This series of three studies examines whether elite collegiate women’s Lacrosse players with GJH may exhibit strength differences that drive the difference in injury risk (Chapter 2), whether there are differences in landing mechanics that drive this difference (Chapter 3), and whether the differences in landing mechanics noted in Chapter 3 were a result of Anterior Cruciate Ligament strain protection (Chapter 4). This chapter summarizes key findings of this dissertation and proposes directions for future studies.

The first study (Chapter 2) sought to determine if there were muscle performance differences between female collegiate athletes with GJH and controls. Previously, muscular strength deficits have been tied to both ACL injury risk and individuals in the general population with GJH. It was unclear whether the strength deficits noted in the GJH population were due to disuse, pain, or something inherent to GJH. If strength
deficits are inherent to the GJH population, that alone may explain the increased injury rate noted in this group. The countermovement jump assessment was chosen as the measure for our study, as it combines aspects of strength, power, speed, and overall athletic performance into a single assessment that is commonly performed by elite athletes. Countermovement jumps are routinely utilized to track athlete physical performance and readiness, they are familiar with the maneuver, and in addition to requiring and assessing strength, it can be collected expediently in a way that does not overly burden our study participants outside of their normal routine. There are many different variables that can be assessed in a countermovement jump, and none of them were statistically different between those with GJH and controls. We therefore feel that GJH does not inherently impact strength in a way that alters physical performance or injury risk. The main limitation of these findings is that they are not an isolated assessment of muscular strength of each key muscle group. In the future, it would strengthen the results to perform isolated muscle testing of the quadriceps, hamstrings, and gastrocnemius/soleus in a way that allows assessment of rate-of-force development and peak strength of each athlete for each muscle group. This was not possible in our setting.

The second study (Chapter 3) examined lower extremity biomechanics during two types of athletic-like tasks to identify if athletes with GJH demonstrated alterations in patterns that may affect knee injury risk. A drop jump is commonly used in assessing lower extremity biomechanics in ACL studies and served as our first assessment. The second task was a more novel cut task – this involved striding forward off of a box onto a single leg, then cutting to the opposite side. This was a strenuous task that required
eccentric muscle activity to land, required balance to land on one leg, and required concentric strength to push off to the side. This was created to simulate an athletic plant and cut maneuver similar to the stresses in athletic competition in a way that was both controllable and reproducible consistently in the lab. Participants quickly grew comfortable and consistent with the task and performed it well. Interestingly, the differences noted in lower extremity biomechanics in the GJH group were identified only during the more strenuous single leg task, and only in the dominant leg. This is not the first study to identify differences between dominant and non-dominant lower extremities in the GJH population (Hanzlikova et al., 2021). Lacrosse would appear to be a fairly symmetric sport with respect to demands on the lower extremity, unlike soccer where there may be a preference for kicking the ball with one leg or the other. However, the “handedness” when handling the Lacrosse stick with the upper extremities may contribute to differences in demands and function of the lower extremities. The differences identified in the dominant leg of the GJH group during the CUT task were consistent with differences generally accepted to increase ACL injury risk. Landing in a more extended hip and knee position has been extensively documented as a component of ACL injuries. Landing with a straighter knee increases the joint compression from the ground reaction force (GRF) through the knee, which is itself a risk factor in ACL injuries. Furthermore, landing on an extended or straighter knee puts the more anterior portion of the femoral articular surface in contact with the tibia, where the shape of the joint surface is flatter versus the more posterior portion of the femur, which is rounder. That flatter surface contacting the tibia is more likely to create a posterior slide of the femur on the tibia, resulting in ACL injury. When the knee is more flexed, that is less
likely because of the shape of the tibia and because the compressive forces across the joint are lower. Shifting loads distally, such as we noted with increased plantar flexor moments in the GJH group has also been associated with females and increased ACL injury risk. Together, this may represent a mechanical reason for the increased knee injury rate in those with GJH.

The reasons for the differences in landing mechanics noted in Chapter 3 are unknown. The third study (Chapter 4) utilized a musculoskeletal model to examine the Anterior Cruciate Ligament strain during the task differences noted in Chapter 3, to assess whether the ACL strain was a factor in guiding lower extremity biomechanics in those with GJH. It was currently not possible in this cohort for us to measure ACL strain directly. We thus utilized a common musculoskeletal model to simulate the ACL strain during the CUT task to analyze whether differences in strain may drive the motor control of the lower extremity. We hypothesized that individuals with GJH may control the knee in a way that minimizes the ACL strain and that may account for the differences in knee mechanics between groups. This was not statistically the case, however. There was no difference across the ground contact phase between the GJH group and controls. While the ensemble averages for ACL length appear visually to slightly differ, the effect size of those differences is less than 1 (d=0.8) and is not detectable with the current study power. It is possible that ACL strain has a small impact on lower extremity mechanics, but it is not something the data here support at this time.

There are limitations to the approach in this group of studies. All studies were done utilizing the same group of athletes. As the studies progressed, injuries were sustained, and not all athletes that performed the initial countermovement jump study
were available for subsequent studies 2 and 3. All subjects came from one United States collegiate Division 1 women’s Lacrosse team. This limits generalizability greatly beyond the particular group studied. On the other hand, this approach and situation greatly reduces the number of extraneous and confounding variables in a way that is not typically possible. The athletes live in the same climate, do the same workouts, practices, with data collected at the same point in the season on the same days. Their daily task demands and sleep/wake schedules are roughly similar, and they are competing at the same level competitively. For all the limitations of a small sample size and homogenous population, there are some very beneficial aspects to these data when examining a complex phenomenon like GJH. For these reasons, this is a step forward and unique contribution to what we know about GJH.

In summary, high level female athletes with GJH demonstrate differences in single-leg land and cut mechanics on their dominant leg. These differences are in patterns associated with increased ACL injury risk. These differences do not appear to be a result of differences in strength or performance inherent to GJH, nor to an attempt to control ACL ligament stress during lower extremity land and cut tasks.
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APPENDICES

Appendix 1

Countermovement jump Box and Whisker plots for variables in Chapter 2. The box represents the range between first and third quartiles, with a line representing the median value dividing the box. An “X” represents the mean value. The lines extending from the box represent the maximum and minimum values in the data. A dot outside of these values represents an outlier value – beyond the 100th or 0 percentile.

![Box and Whisker plots](image-url)
Appendix 2

SPM graphs for variables in Chapter 3.

**Appendix 2.1: SPM of the DJ task Sagittal Plane Angle**
Appendix 2.2: SPM of the DJ task Sagittal Plane Moment
Appendix 2.3: SPM of the DJ task Frontal Plane Angle
Appendix 2.4: SPM of the DJ task Frontal Plane Moment
Appendix 2.5: SPM of the Cut task Sagittal Plane Angle
Appendix 2.6: SPM of the Cut task Sagittal Plane Moment
Appendix 2.7: SPM of the Cut task Frontal Plane Angle
Appendix 2.8: SPM of the Cut task Frontal Plane Moment