DISCOMFORT ASSOCIATED WITH BEYOND END-RANGE ANKLE INVERSION IN HYPERMOBILE AND NON-HYPERMOBILE COLLEGIATE ATHLETES

A THESIS

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by
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THESIS APPROVAL

Graduate Athletic Training Education

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INTRODUCTION

The general population is very rarely diagnosed with a form of joint hypermobility, whether it be Joint Hypermobility Syndrome (JHS) or Ehlers-Danlos syndrome. Both are connective tissue disorders that usually target collagen and cause joint instability, among other connective tissue effects such as easy bruising and increased elasticity in the skin.[1] The joint instability associated with hypermobility can be detrimental to a person's body making them more susceptible to injury due to increased ligamentous laxity. At the same time, persons with a hypermobility syndrome may go through life with no diagnosed injuries, but may complain of generalized pains that seem to have no origin. Conversely, those with an increased ligamentous laxity, athletes especially, may actually be incurring various joint injuries without knowing such as lateral ankle, wrist, or knee sprains. An athlete may regularly "roll" their ankle due to the laxity of the ligaments, but not have pain, hence they do not seek an evaluation by their athletic trainer or physician.

The purpose of this study will be to investigate the difference of discomfort levels felt in association with inversion ankle sprains in hypermobile and non-hypermobile athletes. A significant difference in pain levels could explain a possible lack of diagnosed injuries of athletes with hypermobility and ligamentous laxity. There is a paucity of research on this topic; however, more academic research could significantly help athletes with injury detection and prevention. The results of this study may increase the number of people diagnosed with a hypermobility syndrome via a Beighton Score and in turn help them with how they participate in sports and general daily living activities with the knowledge that they are possibly more susceptible to subconscious injury.

A secondary purpose of this paper is to educate athletic trainers, and other health care professionals, about hypermobility. Athletic trainers that implement a quick Beighton Score screening at the beginning of the season for all athletes could pinpoint those that need individualized prophylactic or injury rehabilitations, and could possibly reduce the number of overall injuries.

METHODS

The purpose of this quasi-experimental study is to investigate if there is a difference in the visual analogue scale score between non-hypermobile and hypermobile athletes when experiencing beyond end-range ankle inversion. The results from this research may indicate if hypermobile athletes injure themselves without feeling pain, or are not injuring themselves in general due to the extensive laxity of their ligaments. This section includes research design, subjects, instrumentation, procedures, hypotheses, and data analysis.

Research Design

This research is a quasi-experimental design. The independent variable is passive ankle inversion range of motion manipulated beyond the athlete's end-range. The dependent variable is the visual analogue scale score which will be measured using a basic 10 cm, 0-10 visual analogue scale (Appendix C5).

Subjects

Participants will be volunteers from the university and club sports teams at California University of Pennsylvania. All university and club hockey athletes will be screened by their assigned graduate assistant athletic trainer using the Beighton Score (Appendix C4) to determine hypermobility status. Those who score a 5/9 or above on the Beighton Score will be considered hypermobile[2] and will be encouraged to volunteer for the study. Those who score a 4/9 or below can volunteer for the study as a non-hypermobile[2] group participant.

Inclusion criteria:

- Between the ages of 18-25.
- Both genders.
- All university sports - football, soccer, golf, swimming, volleyball, cross country, track and field, tennis, basketball, baseball, and softball.
- Club sports - hockey.

Exclusion criteria:

- Any diagnosed ankle injury sustained by the athlete prior to Beighton Score testing for the study.

Instruments

Beighton Score[2]: Appendix C4

The Beighton Score is a nine-part test that includes:

- Passive dorsiflexion of the fifth metacarpophalangeal joint to ≥ 90 degrees on each hand.
- Passive hyperextension of the elbow ≥ 10 degrees for both elbows.
- Passive hyperextension of the knee ≥ 10 degrees for both knees.
- Passive apposition of the thumb to the flexor side of the forearm, while shoulder is flexed 90 degrees, elbow is extended, and hand is pronated for both thumbs.
- Forward flexion of the trunk, with the knees straight, so that the hand palms rest easily on the floor.

Each individual body part is considered a point for a total of nine possible points. A score of 4/9 and below is considered non-hypermobile and a score of 5/9 and above is considered hypermobile.

Handheld Goniometer: Appendix C5

A plastic or metal instrument of range of motion measurement, in degrees, consisting of an axis of rotation, a stationary arm, and a motion arm.

Visual Analogue Scale: Appendix C6

A line, 10cm long, with hash marks and associated numbers at every centimeter and two distinguishing ends. The left end will read "None", the middle will read "Moderate", and the right end will read "Severe".

Procedures

On-campus certified graduate assistant athletic trainers will go over the Beighton Score screening procedure with the research certified graduate assistant athletic trainer. Following certified graduate assistant athletic trainer education, and IRB approval, all university and club hockey athletes will have the informed consent paperwork reviewed with them. Also, they will be informed of the process of the Beighton Score screening, the difference between hypermobile and non-hypermobile, and the definition of ankle inversion and it's range of motion. If they agree to participate, they will sign the informed consent form and be asked their previous ankle injury history. If they have ever had a diagnosed ankle injury, they will be excluded from the study. If they have never had a diagnosed ankle injury, they will then be screened by their certified graduate assistant athletic trainer using the Beighton Score to determine whether or not they fall into the hypermobile or non-hypermobile group. A score 5/9 or higher is considered hypermobile.[2] Those that score high enough to be considered hypermobile will be offered to participate in the study first. Those that score in the non-hypermobile range will be offered to participate after the hypermobile group number is finalized to allow for data collected to be comparable. Once enrolled, participants will be asked to report to the Convocation athletic training facility's doctor's office for one private test session that will last approximately thirty minutes. During the session, the participant will have their passive and active ankle inversion recorded with a goniometer using the tarsal method (range of motion assessment taken with the axis of rotation of the goniometer on the superior border of the talus, the stationary arm of the goniometer aligned with the shaft of the tibia, and the motion arm of the goniometer aligned with the second metatarsal) three times with one minute of rest between measurements. An average range of motion will be determined from the three trials. After their average range of motion is calculated, the certified graduate assistant athletic trainer will read a printed script that states what is expected of the participant. The script reads as follows: "Now that I have an average for your ankle inversion end-range, I will be placing you into beyond that end range. This means that I will start the test at neutral, like the first two sets of tests, and take you to your calculated average end-range based off of the second set of tests. I will let you know when I've reached your average end range and then I will very slowly place you beyond that end range. I need you to tell me when the sensation you feel in your ankle is beyond anything that you consider normal discomfort. For example, when you stretch out in the morning, it feels like a stretch, but it shouldn't be uncomfortable. And then there's other times when you accidentally over stretch and you feel a sensation that isn't normal for you that makes you stop the motion and go back to where you were. When you get to that point, I need you to tell me immediately and I will take a mental note of what degree you got to, place you slowly back into neutral, and then write down the degree. While I record the degree, I need you to circle a number on the visual analogue scale that I will hand you prior to each test. Zero is no discomfort at all and ten is the worst discomfort that you've ever felt in your life and you need me to call the hospital. I would also like you to write down what kind of sensation you felt. Describe it in the best way you know how, no matter how odd it may seem. Remember that you are paying attention for any discomfort that is beyond what you consider normal." Upon understanding of their expectations, they will be passively placed into beyond end-range ankle inversion one degree at a time until they request to end the trial due to abnormal discomfort. The level of discomfort will be recorded by circling the appropriate number on individual visual analogue scales after the participant has been placed back into neutral. This will be performed three times with five minutes of rest between each trial.

Hypothesis

Hypothesis: Hypermobile athletes will report lower visual analogue scale scores with beyond end-range ankle inversion compared to the non-hypermobile athletes.

Data Analysis

Data will be analyzed using SPSS 23.0 with an alpha level of < 0.05. A T-Test will be performed to compare the mean VAS score between hypermobile and non-hypermobile athletes along with the means of the active, passive, and beyond end ranges of motion.

RESULTS

The purpose of this study was to find if there is a difference in discomfort felt between hypermobile and non-hypermobile athletes during beyond-end range ankle inversion. The following section contains pilot data collected for five subjects and is organized in two sections: Demographic Information and Hypothesis Testing.

Demographic Information

There were five total participants in this study; two hypermobile athletes and three non-hypermobile athletes. There were two females and three males with an average age of 20.8 ± .45 years old. This information is displayed in Table 1.

Table 1. Participant Demographics

	Male	Female	Age
Hypermobile	1	1	20.5 ± 0.71
Non-Hypermobile	2	1	21 ± 0

Hypothesis Testing

The following hypothesis was tested for this study with a level of significance set at $\alpha \leq 0.05$.

Hypothesis: Hypermobile athletes will report lower visual analogue scale scores with beyond end-range ankle inversion compared to the non-hypermobile athletes.

Conclusion: Independent-samples *t*-tests were calculated comparing the mean score of active ankle inversion range of motion, passive ankle inversion range of motion, beyond-end ankle inversion range of motion, and the visual analogue scale scores between hypermobile and non-hypermobile participants. No significant difference was found for active ankle inversion range of motion ($t(3)=-.603$, $p>0.05$), passive ankle inversion range of motion ($t(3)=-.467$, $p>0.05$), beyond-end ankle inversion range of motion ($t(3)=-.321$, $p>0.05$), and visual analogue scale scores ($t(3)=-.651$, $p>0.05$). Table 2 shows the means of the three ranges of motion (active, passive, and beyond end-range).

Table 2. Ankle Inversion Range of Motion Means

	Beighton Score Status	Number of Participants	Mean
Active ROM+ (degrees)	Hypermobile	2	31.67° ± 2.33°
	Non-Hypermobile	3	35.89° ± 5.26°
Passive ROM (degrees)	Hypermobile	2	34.17° ± 2.17°
	Non-Hypermobile	3	37.22° ± 4.92°
Beyond End ROM (degrees)	Hypermobile	2	39.00° ± 2.33°
	Non-Hypermobile	3	40.89° ± 4.35°

+ROM=Range of Motion

There was a larger increase in mean inversion range of motion for the hypermobile group from test to test than there was for the non-hypermobile group. The hypermobile group had a 2.5° mean increase from active to passive inversion range of motion while the non-hypermobile group only had a 1.33° mean increase. From passive inversion to beyond end-range inversion, the hypermobile group showed a 4.83° mean increase in range of motion while the non-hypermobile group only showed a 3.67° mean increase in range of motion. Table 3 shows the means of the visual analogue scale scores.

Table 3. Visual Analogue Scale Score Means

	Beighton Score Status	Number of Participants	Mean
VAS* Mean (0-10/10)	Hypermobile	2	0.67/10 ± 0.33/10
	Non-Hypermobile	3	1.33/10 ± 0.78/10

*VAS=Visual Analogue Scale

DISCUSSION

The purpose of this research was to find if there is a difference in discomfort felt between hypermobile and non-hypermobile athletes during beyond-end range ankle inversion to determine if hypermobile athletes may injure themselves without feeling discomfort. The following section is divided into three subsections: Discussion of Results, Conclusions, and Recommendations.

Discussion of Results

To the knowledge of the researcher, this is the first study to compare discomfort felt during beyond end-range ankle inversion between hypermobile and non-hypermobile athletes, or comparing discomfort felt during any form of range of motion between hypermobile and non-hypermobile persons. There is limited research comparing hypermobile and non-hypermobile athletes in any other situation other than gait differences.[3] What other limited research there is focuses on the incidence of injury between the two groups, with opposing, non-significant results. However, most of the research is incomparable due to the vast amount of hypermobility types with some research focused on generalized joint hypermobility and others focused specifically on joint hypermobility syndrome (JHS) or Ehlers-Danlos Syndrome. The aforementioned conditions (JHS and EDS) pertain to hypermobile joints, but with a fundamental difference in that the first is asymptomatic while the others are symptomatic.[3] The lack of evidence is seemingly in line with the results of a study looking at United States' physical therapist's knowledge about joint hypermobility syndrome compared to their knowledge of fibromyalgia and rheumatoid arthritis.[4] The study showed that 38.8% (n=436) were unfamiliar with the diagnosis of JHS and that 73.2% were unfamiliar with the diagnosing criteria.[4] When asked "which conditions are each of the following clinical tests most appropriate", 53.7% of the physical therapists thought that the Beighton Score was not appropriate for assessing JHS, or any of the other three conditions (fibromyalgia syndrome, juvenile rheumatoid arthritis, and adult rheumatoid arthritis) that they named in the study.[4]

The independent-samples *t* tests done for all ranges of motion and the visual analogue scale scores in the current study found no significant results, which seems to be a common trend among the small amount of literature available. The non-hypermobile group demonstrated higher inversion range of motion averages for all three tests with higher standard deviations than the hypermobile group. This means that even though the means were higher within the non-hypermobile group, their averages fell into a wider degree range than the hypermobile group, which were much more clustered together.

Overall, the active range of motion means for both groups were on the higher end of the established norms for ankle inversion as set by the American Academy of Orthopaedic Surgeons (AAOS), which are 0°-35°.[5] The higher active range of motion means, even for the non-hypermobile group, this may be due to the postulation that athletes stretch on a more regular schedule than an average person. With higher active range of motion means, the passive range of motion means would inherently be higher as well, with the participant passive range of motion means being on the very cusp of the average range, or beyond it. This leads to the beyond end-range inversion range of motion means being above the end of the AAOS average range.

The hypermobile group had lower visual analogue scale (VAS)score means, which equated to less discomfort felt during beyond end-range ankle inversion, and their standard deviations, again, were smaller than that of the non-hypermobile group. This showed that the hypermobile group felt less discomfort than the non-hypermobile group. This also suggests that the participants of the hypermobile group had Generalized Joint Hypermobility, rather than Hypermobility Syndrome or Ehlers-Danlos, due to its asymptomatic nature.[3] However, given that the visual analogue scale ran from 0-10 and the means were 0.67 and 1.33 out of ten for the hypermobile and non-hypermobile groups, respectively, a very small amount of discomfort was felt, if any, by all five participants.

Conclusions

Due to a very low sample size, we cannot draw conclusions on the results of this study. Results did show that the hypermobile group, which was made up of two athletes, had lower visual analogue scale score means and that their group showed a larger increase in mean degrees of motion between the three range of motion tests performed. This tells us that they felt less discomfort and that their individual inversion range of motion means were closer together during the testing process.

The non-hypermobile group, which was made up of three athletes, had higher inversion range of motion means for all three tests along with higher standard deviations meaning that their individual means were more widespread than that of the hypermobile group. Their visual analogue scale score means were also higher with a higher standard deviation than the hypermobile group, suggesting that they felt more discomfort.

Even with inconclusive results, it is the thought of the researcher that implementing a quick Beighton Score screening at the beginning of the season for all athletes will help with individualized prophylactic and injury rehabilitations, and possibly reduce the number of overall injuries.

Recommendations

A larger number of participants should be tested in order to have viable results. Multiple schools may need to be included due to the exclusion criteria, imposed by the IRB, which did not take into account the nature of athletics and the injuries that are associated with continually participating in sport from a young age. An electronic goniometer or arthrometer are also suggested to reduce human error.

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APPENDICES

APPENDIX A

Review of Literature

REVIEW OF LITERATURE

This literature review will include three subsections pertaining to multiple components of this study. The anatomy of the ankle will first be reviewed, including composition of ligamentous tissue. Next, there will be an overview of the range of motions of the ankle. Last will be a summary of two hypermobility syndromes and how they are determined.

Ankle Anatomy

The foot and ankle are complex anatomical structures. Their functions include transmitting force between the lower limb and the ground, being a flexible shock absorber, and allowing stable stance and ambulation of the body.[1]

Bony Anatomy

The bony anatomy of the talocrural joint, a true hinge joint, consists of the distal tibial and fibular heads (medial and lateral malleoli, respectively) and the talus.[2,3] The tibia is the primary weight-bearing bone of the leg and has a slightly concaved distal articular surface that forms the roof of the ankle mortise.[3] The fibula is responsible for little to no (0-12%) weight-bearing of the leg, but provides lateral stability to the ankle mortise.[3] The fibula serves as a pulley to increase the effectiveness of the muscles that run behind it and is also a ligamentous attachment site for the anterior talofibular, calcaneofibular, and posterior talofibular ligaments.[3] The talus, which sits under the articular mortise created by the tibia and fibula, is the boundary between the foot and ankle.[3] The saddle shape of the talus is required by the five different functional articulations of which it is a part.[3]

Muscular Anatomy

A combination of muscles and tendons from the anterior, deep posterior and lateral compartments are utilized during uniplanar inversion of the foot. They include the extensor hallucis longus, flexor digitorum longus, flexor hallucis longus, tibialis anterior, tibialis posterior, and the peroneal tendons.[3] The extensor hallucis longus originates from the middle half of the anterior side of the fibula and the interosseous membrane and inserts at the base of the distal phalanx of the great toe.[4] The extensor hallucis longus assists with dorsiflexion and supination.[3]

The flexor digitorum longus originates on the posterior medial portion of the distal two-thirds of the tibia and from the fascia arising from the tibialis posterior and inserts on the bases of the distal phalanges of the second through fifth toes.[3, 4] The flexor digitorum longus assists with ankle plantar flexion and subtalar joint and midtarsal supination.[3] The flexor hallucis longus originates on the lower two-thirds of the posterior surface of the shaft of the fibula along with the associated interosseous membrane and posterior intermuscular septum and inserts on the base of the distal phalanx of the great toe.[4] The flexor hallucis longus assists with the same actions as the flexor digitorum longus.[3]

The tibialis anterior originates on the lateral tibial condyle, the upper one-half of the lateral surface of the tibia, and the adjacent portion of the interosseous membrane and inserts on the medial cuneiform and the base of the first metatarsal.[4] The tibialis anterior assists with ankle dorsiflexion and subtalar joint and midtarsal supination.[3] The tibialis posterior originates on the lateral part of the posterior surface of the tibia, the interosseous membrane, and the proximal half of the posterior surface of the fibula and inserts via fibrous slips at the navicular tuberosity, sustentaculum tali, cuneiforms, cuboid, and the bases of the second through fourth metatarsals.[3, 4] The tibialis posterior assists with ankle plantar flexion and subtalar and midtarsal supination.[3]

The peroneal muscles consist of three muscles, whose brevis and longus tendons run behind the lateral malleolus.[3] The peroneus brevis originates on the lower two-thirds of the lateral surface of the fibula and inserts on the lateral side of the base of the fifth metatarsal.[4] The peroneus brevis assists with subtalar joint and midtarsal pronation and ankle plantar flexion.[3] The peroneus longus originates on the lateral tibial condyle, fibular head, and the upper two-thirds of the lateral surface of the fibula and inserts on the lateral side of the medial cuneiform and the base of the first metatarsal.[4] The peroneus longus assists with subtalar joint and midtarsal pronation and ankle plantar flexion.[3] The peroneus tertius originates on the lower third of the anterior surface of the fibula and the interosseous membrane and inserts on the dorsal surface of the base of the fifth metatarsal.[4] The peroneus tertius assists with subtalar joint and midtarsal pronation and ankle dorsiflexion.[3]

Ligamentous Anatomy

The lateral collateral ligament complex of the ankle is made up of the anterior talofibular, calcaneofibular, and posterior talofibular ligaments which are all thickenings of the lateral capsule of the ankle.[5, 6] The anterior talofibular ligament (ATF), which is a primary stabilizer of the ankle preventing forward subluxation of the talus, can have anywhere from one to three bands (but usually two), originates at the anterior margin of the lateral malleolus and runs anteromedially to its insertion on the talar body immediately anterior to the joint surface occupied by the lateral malleolus.[6, 7, 8] The ATF is essentially parallel to the ankle when the ankle is in a neutral position.[6] When the ankle is in dorsiflexion, the ATF moves upward with the inferior band becoming taut and the superior band remaining lax.[6] When the ankle is in plantar flexion, the ATF moves downward with the superior band becoming taut and the inferior band remaining lax.[6] Plantar flexion is the anterior talofibular ligament's most susceptible position to injury, especially when the ankle is also inverted.[6]

The calcaneofibular ligament (CF), which is the primary restraint of talar inversion, originates at the anterior portion of the lateral malleolus and runs diagonally, inferiorly and posteriorly, to its insertion on the posterior region of the lateral calcaneal surface.[6, 8] The CF becomes horizontal during ankle extension and vertical during ankle flexion while remaining taut throughout the entire range of motion, whereas the CF is lax in a neutral, valgus position and taut in a relaxed, varus position.[6] Even with the CF being taut more often than not, isolated injury to the calcaneofibular ligament is extremely rare.[6]

The posterior talofibular ligament (PTF) originates at the malleolar fossa on the medial surface of the lateral malleolus and runs horizontally to its insertion on the posterolateral talus at varying locations including the posterior surface of the talus, the os trigonum (if present), possibly contributing to forming the tunnel for the flexor hallucis longus tendon, and fusing with the posterior intermalleolar ligament.[6] The PTF is lax in neutral and plantar flexed positions while taut in dorsiflexion.[6] Even with being taut in dorsiflexion, the PTF is usually not injured unless there is a "frank" dislocation of the ankle.[6]

Ligaments are made of mostly Type I, with some Type III, collagen fibers which are found of fibril form in dense fibrous connective tissue.[9] The mechanical stability found in Type I collagen of ligaments is due to the rod-like shape of the collagen and its inherent flexibility.[9] Genetic disorders via gene variants, such as Ehlers-Danlos, can affect certain types of collagen and cause hypermobility disorders.[10]

Ankle Range of Motion

The uniplanar motions of the talocrural joint, which is a true hinge joint, are only dorsiflexion and plantar flexion, while the subtalar joint motions include inversion/eversion and abduction/adduction.[2, 3, 11] Average tarsal measurement range of motion for inversion is between 0° and 35°, while eversion is between 0° and 15° (Table 4).[12] Average range of motion for dorsiflexion is anywhere from 0°-20°, while plantar flexion is between 0° and 50°.[12]

Table 4. Average Ankle Ranges of Motion[12]

Motion	Average Range of Motion (°)
Dorsiflexion	0° - 20°
Plantar Flexion	0° - 50°
Inversion	0° - 35°
Eversion	0° - 15°

The motions around the oblique axis of the ankle combine the uniplanar motions together to make up pronation (dorsiflexion, abduction, and eversion) and supination (plantar flexion, adduction, and inversion).[3] Closed-chain pronation causes internal tibial rotation, knee flexion, and internal rotation of the hip, while closed-chain supination causes the opposite pronation with external tibial rotation, knee extension, and external rotation of the hip.[3]

The extensor hallucis longus, flexor digitorum longus, flexor hallucis longus, tibialis anterior, and tibialis posterior all help with ankle inversion.[3] During inversion the anterior talofibular ligament, calcaneofibular ligament, posterior talofibular ligament, lateral capsule, and peroneal tendons are all subject to tensile forces while the deltoid ligament is subject to compressive forces.[3]

Joint Hypermobility

Joint hypermobility is often defined as a connective tissue disorder that causes an extreme range of joint play in the synovial joints.[13] Joint hypermobility is considered a hypermobility syndrome when there are symptoms accompanying the disproportionate joint play.[13] Eighty percent of survey responding physiotherapists in the UK stated that they think that hypermobility syndrome has a significant impact on the patient's quality of life.[14]

Joint Hypermobility Syndrome

Joint Hypermobility Syndrome (JHS) has been classified as a hereditary connective tissue disorder, associated with excessive joint range of motion in the presence of pain, much like Marfan's Syndrome and Ehlers-Danlos hypermobility type.[13, 15] Benign Joint Hypermobility Syndrome is joint hypermobility, and other musculoskeletal problems, which can be acquired through years of excessive stretching, such as with gymnasts or ballet dancers.[13, 16] While a very small amount of the population is formally diagnosed, a surprisingly large portion of the population suffers from JHS.[13] Research shows that there is a difference in prevalence of JHS when comparing sex, race, and age with young, non-Caucasian females having the highest incidence of the hypermobility syndrome phenotype.[13] JHS is often attributed to an atypical ratio of type III to type I collagen fibers.[13] It does not affect just the collagen and the range of joint play; it also has nervous system effects, such as proprioceptive acuity and altered neuromuscular reflexes, which are probable causes of predisposition to injury and tissue damage.[13]

In some cases joint hypermobility can be considered an asset and not result in problems, but that is generally not the case.[13] The tissue laxity associated with JHS can be the root of a variety of debilitating symptoms such as persistent, longstanding pain, joint stiffness, an assortment of sounds (clicking, popping, clunking, etc.), sublux/dislocations and instability, and feeling that joints are vulnerable or feeling like a '90 year old'.[13] Other JHS symptoms consist of numbness, tiredness, faintness, not feeling well, and flu-like symptoms.[13] JHS has been reported to present with extra articular manifestations including skin laxity, autonomic disturbances, eyelid drooping, varicose veins, bruising, Raynaud's phenomenon, developmental motor coordination delay, neuropathies, tarsal and carpal tunnel syndromes, fibromyalgia, low bone density, anxiety and panic states, and depression.[13] With the wide range of symptoms listed above as compared to the way that the patient looks or moves, often times those suffering from JHS are misunderstood and taken for hypochondriacs and even labeled as possibly having psychological problems.[13] Eighty percent of survey responding physiotherapists in the UK stated that they think that JHS has a significant impact on the patient's quality of life.[14] Healthcare professionals need to be sure to look at the patient as a whole and not just the singular problem area if JHS is a possible diagnosis for the patient's list of symptoms. The inclusion of a Beighton Score during the assessment of a new patient can prove to be invaluable in diagnosing JHS and making sense of the patient's current symptoms.

Ehlers-Danlos Syndrome

Ehlers-Danlos Syndrome (EDS) is very similar to Hypermobility Syndrome concerning presentations and symptoms with the main difference being that the majority of the different types of EDS have distinct genetic markers and very specific manifestations.[17] There are three major and three minor EDS types: The major types are Classical, Hypermobile, and Vascular.[17] The minor types are Kyphoscoliosis, Arthrochalasia, and Dermatosparaxis.[17] The Hypermobile type is the most prevalent and can be represented by generalized hypermobility of large and small joints all over the body, a score of 5/9 or higher on the Beighton Score, and debilitating chronic limb and joint pain even with normal image findings.[17] Hypermobile type EDS has an autosomal dominant transmission through families and can be diagnosed partly with a family history.[17]

Beighton Score

The Beighton Score is a nine point test consisting of five tests with four of the tests having a bilateral score.[18] The first four tests are looking as specific, bilateral passive ranges of motion for a particular joint.[18] If only one side reaches a positive range of motion, there is only one point awarded for the test, if both sides are positive, two points are awarded.[18] The first test is looking at passive dorsiflexion of the fifth metacarpophalangeal joint on both hands.[18] A positive for this test would be dorsiflexion $\geq 90°$.[18] The second test is looking at passive hyperextension of the elbows.[18] A positive for test two would be hyperextension $\geq 10°$.[18] The third test looks at passive hyperextension of the knees.[18] This test is positive if the hyperextension is $\geq 10°$.[18] The fourth test looks at passive apposition of the thumb to the flexor side of the forearm while the shoulder is flexed to 90°, the elbow is extended, and the hand is pronated.[18] A positive for this test would be if the whole thumb touches the flexor side of the forearm.[18] The fifth test looks at forward flexion of the trunk with knees straight.[18] A positive for this test would be if the palms of the hands rest easily on the floor, counting for a single point.[18]

Table 5. Beighton Score[18]

Description	Bilateral Testing	Scoring (max. points)
Passive dorsiflexion of the fifth metacarpophalangeal joint to ≥ 90 degrees	Yes	2
Passive hyperextension of the elbow ≥ 10 degrees	Yes	2
Passive hyperextension of the knee ≥ 10 degrees	Yes	2
Passive apposition of the thumb to the flexor side of the forearm, while shoulder is flexed 90 degrees, elbow is extended, and hand is pronated	Yes	2
Forward flexion of the trunk, with the knees straight, so that the hand palms rest easily on the floor	No	1
Total		9

This scale is widely used due to its validity but can garner very different results depending on what number out of nine the researcher sets for their inclusion criteria. For adults it is usually set to greater than, or equal to, 4/9, but if a researcher were to use that number for their inclusion criteria for children, the number of hypermobile participants will be much higher than if it were set to a more reasonable greater than, or equal to 6/9, since children are inherently more flexible than adults.[19]

Summary

The bones of the ankle consist of the distal ends of both the tibia and fibula as well as the talus. The majority of the muscles from the anterior, deep posterior, and lateral compartments of the lower leg are utilized during uniplanar inversion of the ankle. The lateral collateral complex of the ankle, utilized mainly for stability, includes the anterior talofibular, calcaneofibular, and posterior talofibular ligaments which are all thickenings of the lateral capsule of the ankle. Joint hypermobility is often defined as a connective tissue disorder, such as Hypermobility Syndrome or Ehlers-Danlos Syndrome, that causes an extreme range of joint play in the synovial joints, especially in the ankle. The Beighton Score is a five test assessment that checks the range of motion to see if a particular patient has joint hypermobility.

APPENDIX B

The Problem

STATEMENT OF THE PROBLEM

It is important to study the relationship of discomfort levels between hypermobile and non-hypermobile athletes. A large portion of the athletic population is actually hypermobile but has never been diagnosed or even checked for a hypermobility syndrome. If there is a lack of discomfort felt with hypermobile athletes during beyond end-range ankle inversion this may lead researchers to assume that hypermobile athletes are actually injuring themselves without feeling associated discomfort. This knowledge could lead to increased awareness and care for hypermobile athletes on all levels. The purpose of this study is to examine if a relationship between discomfort levels during beyond end-range ankle inversion with hypermobile and non-hypermobile athletes exists.

Definition of Terms

The following definitions of terms will be defined for this study:

1. Ankle inversion range of motion - an average normal range of 0°-35° using a tarsal measurement.[12]

2. Tarsal measurement method - range of motion assessment taken with the axis of rotation of the goniometer on the superior border of the talus, the stationary arm of the goniometer aligned with the shaft of the tibia, and the motion arm of the goniometer aligned with the second metatarsal.
3. Goniometer - a plastic or metal instrument of range of motion measurement, in degrees, consisting of an axis of rotation, a stationary arm, and a motion arm.
   a. Axis of rotation - the center connection point of the stationary arm and the motion arm that is surrounded by a full protractor that can measure all degrees between 0° and 360°.
   b. Stationary arm - the arm of the goniometer that has the full protractor on it that is held in one place so that the motion arm can move around the protractor to correctly measure the specific joint range of motion.
   c. Motion arm - the arm of the goniometer that moves around the stationary arm and protractor to correctly measure specific joint range of motion.
2. Visual analogue scale/VAS - a line, 10cm long, with hash marks and associated numbers at every centimeter.

The left end will read "None", the middle will read "Moderate", and the right end will read "Severe".

3. Hypermobility/hypermobile - athletes scoring 5/9 or greater on the Beighton Score of Hypermobility.[20] Hypermobile athletes may have an average ankle inversion range of motion higher than the average range.
4. Non-hypermobility/non-hypermobile - athletes scoring a 4/9 or below on the Beighton Score of Hypermobility.[20]
5. Previous history - any diagnosed ankle injury ever experienced by the athlete in their life.

Basic Assumptions

The following are basic assumptions of this study:

1. It is assumed that the athletes participating in this study will give true and honest answers on both their inclusion criteria answers and on the visual analogue scale.
2. It is assumed that the primary researcher using the goniometer will accurately measure ankle inversion each time.

3. It is assumed that age, weight, race, and sport played will have no effect on the outcomes of the visual analogue scale.

Limitations of the Study

The following are possible limitations of the study:

1. The athlete's willingness to fully participate in this study.
2. The athlete's documented level of discomfort as a true representation of the actual discomfort felt.
3. The athlete's level of pain tolerance.

Significance of the Study

This research is being done on a topic with very little published research. There have been very few articles that come close to examining pain and range of motion among hypermobile and non-hypermobile athletes. Joint hypermobility is considered an asymptomatic increased range of joint movement and is about three times more common in females than in males.[20] Most studies examine the prevalence rates of hypermobility syndromes within certain populations, such as dancers or general groups of children.

These studies find that there are correlations between certain sports and hypermobility as well as between gender, age, and population.[21, 22, 23] While there are really no contradictory findings in the research, a majority of articles have come to the conclusion that hypermobility syndromes are grossly under diagnosed whether they be a score from a Beighton Score for generalized joint hypermobility or specific type of Ehlers-Danlos syndrome.[17, 18, 24]

Ankle sprains are the most common type of athletic injury accounting for 10-15%.[25] Ligamentous injuries to the lateral complex of the ankle are the most common form of ankle sprain.[25] Functional instability, such as chronic giving way and increased lateral ligament laxity, of the ankle joint is the most common residual disability following an inversion ankle sprain in 10-41% of patients.[2, 26, 27] Previously injured ankles due to lateral ankle sprains show more inversion rotation and less inversion and anterior stiffness indicating that ligamentous laxity is not the only mechanical tissue characteristic that changes after a lateral ankle sprain.[28] According to one study, generalized joint laxity is not a factor when it comes to ankle ligament injury, while another study states that it has been implicated with ankle sprains.[29, 30]

Comparing discomfort levels in hypermobile and non-hypermobile groups during ranges of motion has not been extensively studied leading to an enormous gap in the research of hypermobility syndromes and injury rates. Studying this relationship could lead to a wealth of information regarding specific joint injury prevalence and healing rates among hypermobile athletes. Are hypermobile athletes injuring themselves and just not registering the discomfort, or are their ligaments too lax to even be injured? Researching this relationship could be the beginning to finding answers to the above questions. It could also lead to an increase in hypermobility diagnoses and awareness among athletic trainers and physicians.

APPENDIX C

Additional Methods

APPENDIX C1

Informed Consent Form

1. Ana Elo, who is a Graduate Athletic Training Student at California University of Pennsylvania, has requested my participation in a research study at California University of Pennsylvania. The title of the research is Discomfort Associated with Beyond End-Range Ankle Inversion in Hypermobile and Non-Hypermobile Collegiate Athletes.

2. I have been informed that the purpose of this study is to determine differences in discomfort felt during beyond end-range ankle inversion between hypermobile and non-hypermobile collegiate athletes. I understand that I must be 18 years of age or older to participate. I understand that I have been asked to participate along with other varsity athletes participating in all university sports as well as club hockey at California University of Pennsylvania. I understand that to participate in data collection I can confirm I have not had an ankle injury prior to the beginning of testing.

3. I have been invited to participate in this research project. My participation is voluntary and I can choose to discontinue my participation at any time without penalty or loss of benefits. I had the opportunity to ask questions about the study. My participation will involve Beighton Score testing, active and passive ankle inversion range of motion, and passive beyond end-range ankle inversion.

First, the participants will go through the Beighton Score for placement into hypermobile and non-hypermobile groups. Once subjects are deemed eligible for participation, all ankle inversion measurements will be taken, privately, during one testing session. All testing will be completed in one day.

4. I understand there are minimal foreseeable risks to me if I agree to participate in the study. I understand that there is risk of pain during testing. With participation in a research program such as this there is always the potential for unforeseeable risks, such as lingering pain. I understand the Principal Investigator will inform me of any significant new findings developed during the research that may affect me and influence my willingness to continue participation.

5. I understand that, in case of injury, I can expect to receive treatment or care in Hamer Hall's Athletic Training Facility. This treatment will be provided by the researcher, Ana Elo, a licensed and certified athletic trainer. Additional services needed for prolonged care will be referred to the attending staff at the Downey Garofola Health Services located on campus and that any medical expenses incurred will be processed though my primary insurance provider.

6. There are no feasible alternative procedures available for this study.

7. I understand that the possible benefits of my participation in the research is the better understanding of discomfort felt by hypermobile athletes and that my participation may aid in the alteration of prevention and treatment techniques to reduce the likelihood of future ankle sprains in those with hypermobility.

8. I understand that the results of the research study may be published but my name or identity will not be revealed. Only aggregate data will be reported. In order to maintain confidentially of my records, Ana Elo will maintain all documents in a secure location on campus and password protect all electronic files so that only the student researcher and research advisor can access the data. Each subject will be given a specific subject number to represent his or her name so as to protect the anonymity of each subject.

9. I have been informed that I will not be compensated for my participation nor will there be any costs incurred on my behalf.

10. I have been informed that any questions I have concerning the research study or my participation in it, before or after my consent, will be answered by:

Ana Elo, LAT, ATC

Graduate Assistant Athletic Trainer/Primary Researcher

ELO2788@calu.edu

(407) 832-5154

Dr. Shelly DiCesaro

Research Advisor

Dicesaro@calu.edu

(724) 938-5831

11. I understand that written responses may be used in quotations for publication but my identity will remain anonymous.

12. I have read the above information and am electing to participate in this study. The nature, demands, risks, and benefits of the project have been explained to me. I knowingly assume the risks involved, and understand that I may withdraw my consent and discontinue participation at any time without penalty or loss of benefit to myself. I understand the Principal Researcher may terminate my participation at any time without warning. In signing this consent form, I am not waiving any legal claims, rights, or

remedies. A copy of this consent form will be given to me upon request.

13. This study has been approved by the California University of Pennsylvania Institutional Review Board.

14. The IRB approval dates for this project are from: 04/17/2016 to 04/18/2017.

Subject's Signature: ______________________ Date: _________

Witness Signature: ________________________ Date: _________

APPENDIX C2

Institutional Review Board –

California University of Pennsylvania

Institutional Review Board

California University of Pennsylvania

Morgan Hall, Room 310

250 University Avenue

California, PA 15419

instreviewboard@calu.edu

Robert Skwarecki, Ph.D., CCC-SLP,Chair

Dear Ms. Elo:

Please consider this email as official notification that your proposal titled "Pain Associated with Beyond End-Range Ankle Inversion in Hypermobile and Non-hypermobile Collegiate Athletes" (Proposal #15-068) has been approved by the California University of Pennsylvania Institutional Review Board as amended with the following stipulations:

This approval extends only to a trial of five (5) participants, at least two (2) of which are classified as "hypermobile" according to the criteria used in the study. Once data has been collected from the 5 participants, the researcher must stop data collection and report initial results to the Board. Data provided to the Board must be reported separately for each participant (identities kept anonymous) and include:

-Range of motion under standard testing;

-Degrees beyond-end-range obtained before the test was stopped;

-Discomfort scale rating;

-Participant description of the sensation

-Any complaints or incidents encountered during the trial.

Upon receipt, the Board will review the initial results. At that point the Board can issue an approval to continue with the full cohort of anticipated participants and/or modifications as appropriate.

Once you have made these changes you may immediately begin data collection. You do not need to wait for further IRB approval. At your earliest convenience, you must forward a copy of the changes for the Board's records.

The effective date of the approval is 04/19/2016 and the expiration date is 04/18/2017. These dates must appear on the consent form.

Please note that Federal Policy requires that you notify the IRB promptly regarding any of the following:

(1) Any additions or changes in procedures you might wish for your study (additions or changes must be approved by the IRB before they are implemented)

(2) Any events that affect the safety or well-being of subjects

(3) Any modifications of your study or other responses that are necessitated by any events reported in (2).

(4) To continue your research beyond the approval expiration date of 04-18-2017 you must file additional information to be considered for continuing review. Please contact instreviewboard@cup.edu. Please notify the Board when data collection is complete.

Regards,

Robert Skwarecki, Ph.D., CCC-SLP

Chair, Institutional Review Board

APPENDIX C3

Individual Data Collection Sheet

Subject #: ________________

Sex: ________________

Age: ________________

Beighton Score

Description	Left	Right
Passive dorsiflexion of the fifth metacarpophalangeal joint to ≥ 90 degrees		
Passive apposition of the thumb to the flexor side of the forearm, while shoulder is flexed 90 degrees, elbow is extended, and hand is pronated		
Passive hyperextension of the elbow ≥ 10 degrees		
Passive hyperextension of the knee ≥ 10 degrees		
Forward flexion of the trunk, with the knees straight, so that the hand palms rest easily on the floor		
Total		

Inversion Range of Motion

	Active ROM	Passive ROM	Beyond End ROM
1			
2			
3			
Average			

APPENDIX C4

Beighton Score

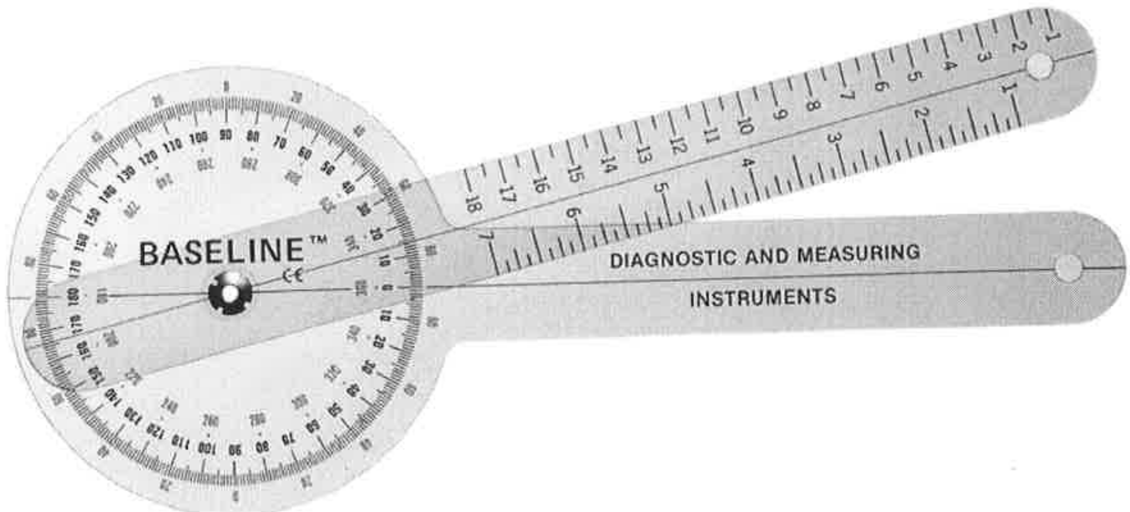
Beighton Score

Description	Bilateral Testing	Scoring (max. points)
Passive dorsiflexion of the fifth metacarpophalangeal joint to ≥ 90 degrees	Yes	2
Passive hyperextension of the elbow ≥ 10 degrees	Yes	2
Passive hyperextension of the knee ≥ 10 degrees	Yes	2
Passive apposition of the thumb to the flexor side of the forearm, while shoulder is flexed 90 degrees, elbow is extended, and hand is pronated	Yes	2
Forward flexion of the trunk, with the knees straight, so that the hand palms rest easily on the floor	No	1
Total		9

APPENDIX C5

Goniometer

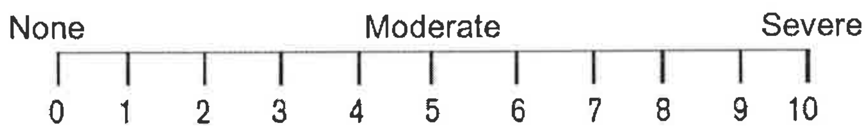
Goniometer



APPENDIX C6

Visual Analogue Scale

Visual Analogue Scale



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ABSTRACT

Background: Hypermobility is widely under diagnosed in all demographic areas, including athletes. Hypermobile athletes could be at a greater risk of unnoticed injury when compared to non-hypermobile athletes. With lateral ankle sprains being the most common athletic injury, this could pose serious problems. Athletic trainers could have hypermobile athletes and mistake their naturally lax ankle ligaments and associated problems with chronic ankle instability if a proper history is not obtained. The purpose of this study is to determine whether or not hypermobile athletes have decreased sensations as compared to non-hypermobile athletes. **Methods:** The Beighton Score was utilized to determine hypermobility status among collegiate athletes with a score of 5/9 or above considered to be hypermobile. Active and passive ankle inversion range of motion (ROM) were taken for all athletes in both the hypermobile and non-hypermobile groups to establish an end range. Athletes were then placed into beyond their end range until discomfort was felt, up to 10° beyond their end range, or a firm end-feel was felt. Athletes then rated their discomfort on a visual analogue scale (VAS).
Subjects: Five participants age 20.8±.4472 years old with 2 hypermobile athletes (1 male, 1 female) and 3 non-

hypermobile athletes (2 males, 1 female). **Results:** No significant results were obtained due to the small number of participants. The non-hypermobile group had higher ROM means and standard deviations (AROM: 35.89°±9.12°, PROM: 37.22°±8.51°, BEROM: 40.89°±7.53°) than the hypermobile group (AROM: 31.67°±3.3°, PROM: 34.17°±3.06°, BEROM: 39°±3.3°) for all three tests, but the hypermobile group had lower VAS score means and standard deviations (.67/10±.47/10) than the non-hypermobile group (1.33/10±.1.33/10). **Conclusions:** Participant numbers are too low to make a statement regarding significant results. The non-hypermobile group had a larger range for their range of ankle inversion, while the hypermobile group has a more compact range of ankle inversion. The VAS scores are also very low for both groups, suggesting very little discomfort, if any, felt by either group.