Viscoelastic Properties and Flexibility of the Human Muscle-Tendon Unit in Benign Joint Hypermobility Syndrome

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ABSTRACT. ***Objective***. To examine the passive energy absorption of the hamstring muscle-tendon unit in 9 women with benign joint hypermobility syndrome (BJHS) and 9 age and sex matched controls.

Methods. Resistance to stretch was measured as knee flexion moment (N·m) with an isokinetic dynamometer during passive knee extension. The angle that induced a stretch sensation without pain was the maximal stretch angle. Force, angle, angular velocity (0.09 rad/s), and electromyograph were simultaneously and continuously recorded during the stretch. Hamstring cross sectional area was obtained with magnetic resonance imaging.

Results. Forearm skin extensibility was greater for BJHS (3.6 ± 0.5 cm) than for controls (1.3 ± 0.2 cm) ($p < 0.01$). Similarly, the Beighton score was greater for BJHS (6.6 ± 0.8) than for controls (0.4 ± 0.2) ($p < 0.001$). Maximal stretch angle was greater for BJHS (Δ1.35 ± 0.07 rad) compared to controls (Δ0.98 ± 0.05 rad) ($p < 0.001$), and the corresponding peak moment was also greater for BJHS (1.3 ± 0.4 Nm/cm^2) than for controls (0.6 ± 0.1 Nm/cm^2) ($p < 0.01$). For a given mutual angle the passive energy absorption was the same for both groups, but at the maximal stretch angle the total area–normalized energy was greater for BJHS (0.36 ± 0.04 J/cm^2) than for controls (0.18 ± 0.28 J/cm^2) ($p < 0.001$).

Conclusion. The lack of difference in passive energy absorption for a given mutual stretch angle suggests that passive properties of the muscle-tendon unit of BJHS are similar to those of controls. However, the greater maximal stretch angle and corresponding peak moment in BJHS suggests a greater subjective tolerance to passive stretch. That is, increased flexibility in BJHS is not a function of altered passive properties of the muscle-tendon complex. It remains unknown if the enhanced tolerance to passive tension plays a role in the development of musculoskeletal ailment. (J Rheumatol 2001;28:2720–5)

Key Indexing Terms:
FLEXIBILITY STRETCH STIFFNESS ENERGY ABSORPTION

There are several heritable disorders of connective tissue that result from alterations in the synthesis or structure of collagen, elastin, or extracellular matrix components[1-3]. One of the connective tissue disorders is the benign joint hypermobility syndrome (BJHS), which has clinical manifestations that vary greatly, and to some extent has considerable overlap with Marfan and Ehlers-Danlos syndromes, and osteogenesis imperfecta[3]. Joint hypermobility is unequivocally the most striking feature of BJHS, although it also encompasses several other non-life-threatening complications, including arthralgia and skin involvement[3,4]. In addition, it is not uncommon for patients with BJHS to suffer orthopedic complications that severely affect their function, including dislocations and joint pain due to the excessive joint motion[4-6]. Hypermobile joints and excessive musculoskeletal flexibility, which may contribute to joint instability, are believed to be related to reduced stiffness of the muscle-tendon unit and the joint capsule in patients with inherited connective tissue disorders[7-9]. Consequently, these patients have been recommended to perform strengthening exercises to increase the joint stability and potentially lessen joint trauma.

The passive properties of the muscle-tendon during stretching can be explained in biomechanical terms[10,11]. The static stretch of a muscle can be divided into a lengthening phase, followed by a static phase in which a constant muscle-tendon length is maintained for some time[11]. The slope of the linear portion of the length-tension curve in the lengthening phase and the area under the curve represent tissue stiffness and energy, respectively, while the decline in moment with time in the static phase represents viscoelastic stress relax-

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ation. Both stiffness and energy absorption, and viscoelastic stress relaxation have recently been described in a human model[12-14]. However, whether the reported excessive range of motion in individuals with BJHS can be attributed to reduced passive mechanical properties of the muscle-tendon and/or the joint capsule remains unknown. We investigated the passive mechanical properties of the muscle-tendon unit in the hamstring muscle in individuals with BJHS compared that to age and sex matched controls.

MATERIAL AND METHODS

Nine female patients with BJHS were recruited from Bispebjerg Hospital Dermatology Department, Copenhagen — age 34.0 ± 6.8 yrs; height 1.66 ± 0.07 m; body mass 62.8 ± 8.1 kg, mean ± SD. They all fulfilled the recently published criteria for BJHS[3]. The Beighton score, which includes (1) hyperextension of the 5th finger, (2) apposition of the thumb, (3) hyperextension of the elbow, (4) hyperextension of the knee, and (5) placing the palms of the hands on the floor with knees extended, was 4/9 or greater for 8/9 subjects, with a mean of 6.6 (range 3–9). Eight of nine subjects had longstanding arthralgia. Four subjects had joint subluxations. Four subjects had easy bruising and one had scarring. All patients had skin biopsies taken for electron microscopy examination, which revealed that 8 subjects had morphological changes in the structure of collagen fibrils (twisting). An age and sex matched group of 9 healthy subjects served as controls (age 32.1 ± 7.6 yrs; height 1.69 ± 0.07 m; body mass 67.6 ± 13.6 kg, mean ± SD).

Instrumentation. Resistance to stretch was defined as the passive moment (N·m) offered by the hamstring muscle group during passive knee extension, measured using a KinCom dynamometer (Kinetic Communicator, Chattecx Corp., Chattanooga, TN, USA) with a modified thigh pad. The measurement technique has been described in detail[12,14,15]. Based on the calculated moment about the knee (N·m) the tensile force on the hamstring muscles can be derived using mathematical modeling with instantaneous muscle length and joint moment arm for each muscle[16]. However, for the purposes of this study the moment was taken to represent the tensile loading of the hamstring muscles. Subjects were seated with the trunk perpendicular to the seat for the stretch procedure. The thigh rested on a specially constructed thigh pad that elevated the thigh to 0.524–0.785 rad above horizontal. The trunk and thigh position did not allow subjects to reach complete knee extension. Consequently, the position of the subject during the stretch maneuver placed tension primarily on the muscle-tendon unit without involvement of posterior capsular constraints about the knee. Passive force (N) was detected by the load cell of the dynamometer, which was calibrated prior to the experiment (10–750 N). The dynamometer and knee joint axis were aligned and the moment (N·m) about the knee joint was calculated by multiplying the measured force by the lever arm distance. The lever arm attachment was placed 2 cm proximal to the lateral malleolus. The distal thigh and pelvis were firmly secured with straps to minimize joint movement during the stretch maneuver. Reliability of the method has been shown to yield a correlation coefficient of r = 0.99 with a coefficient of variation of 5.8–6.5% with respect to passive moment[17].

Gross electrical activity of the human hamstring muscle group was measured with Ag/AgCl surface electrodes (Type N-10-A, Medicotest, Olstykke, Denmark) placed midway between the gluteal fold and the knee joint, with a 3 cm inter-electrode distance. Custom made amplifiers with a frequency response of 20 Hz to 10 kHz and 1:1 preamplifiers were used for electromyography (EMG) signal sampling. The EMG signal was full wave rectified and averaged (time constant 200 ms)[18]. Using an identical measurement technique as in this study we have shown that there is no contractile activity of the hamstring muscle group that contributes to the resistance in the lengthening phase or subsequent viscoelastic stress relaxation during the static phase[12,15,19]. In agreement with previous data, low amplitude EMG recordings showed that the contractile component did not contribute to resistance to stretch in the lengthening or static phase of the stretch[12,15,19].

Stretch maneuver. The stretch maneuver consisted of a lengthening phase to a predetermined final angle followed by a 90 s static phase (Figure 1). The final angle during the stretch maneuver was determined by the examiner passively lifting the lever arm and thereby extending the knee to an angle that provoked a subjective sensation of tightness in the posterior thigh similar to a static stretch maneuver. The determination of the maximal stretch angle served as a preconditioning[20]. It has been reported that only the first of 10 consecutive stretches significantly affects energy, while stiffness remains unaffected[21]. The leg was then immediately returned to the starting position. The dynamometer was programmed to extend the knee passively with an angular velocity of 0.087 rad/s (5°/s) from the starting point of 1.222 rad (70°) below horizontal to the final angle (lengthening phase), where it remained for 90 s (static phase). Positions above horizontal were indicated as negative values. Subjects were requested to relax completely and not offer any voluntary resistance throughout the stretch maneuver.

Muscle cross sectional area. Passive resistance to stretch will depend on both the quality and quantity of tissue in parallel. It has been shown that there is a positive relationship between hamstring cross sectional area and resistance to stretch[14]. That is, the larger muscle will tend to offer more resistance. Therefore, the cross sectional area of the hamstring muscles was determined on a magnetic resonance image (MRI) to allow for normalization of size. The MRI was obtained at 50% of the distance between the greater trochanter and the lateral joint line of the left knee (2D T1 weighted fast field echo) (TR/TE, 500/14 ms; FOV180; matrix 512 × 512; slice thickness 6 mm). The cross sectional area (cm^2), which included biceps femoris long head, biceps femoris short head, semitendinosus, and semimembranosus, was calculated by tracing the borders of these muscles on a computer image. Repeated measures of the cross sectional area yielded a coefficient of determination of $r^2 = 0.99$ and a coefficient of variation of 2.8% for duplicate measures.

Skin extensibility. In addition to the measurement of passive properties of the hamstring muscle-tendon, clinical tests, the Beighton assessment, and the skin extensibility were measured[22]. The skin was measured halfway down the volar surface of the forearm by grasping and pulling the skin out for measurement with a ruler[22].

Data reduction and analysis. Passive force, joint range of motion, angular velocity, and hamstring EMG were continuously recorded for all stretch maneuvers. Signals were sampled at 50 Hz, A/D converted, and stored on a PC for subsequent analysis. A 4th order polynomial fit was applied to the moment-angle curves in the lengthening phase of the stretch maneuvers (Figure 2). Passive energy absorption was calculated as the area covered by the polynomial moment-angle curve in the entire range by converting the angular change into radians. Because the maximal angle during the stretch is individually determined, comparisons of passive energy between the groups were made based on (1) a joint angle common to all subjects (Figure 4), and (2) the subject's maximal joint angle (Figure 5). For this study passive energy during the stretch was defined as muscle area-normalized energy (J/cm^2). Area-normalized peak moment (Nm/cm^2) was obtained the instant the lever arm reached the final angle, and decline in moment was expressed as a percentage of peak and referred to as viscoelastic stress relaxation[12,14,15]. Mann-Whitney U ranks tests were used to determine whether differences existed between BJHS and control subjects for the measured variables. An alpha level of 0.05 was considered significant. Results are reported as mean ± SEM.

RESULTS

Forearm extensibility was greater for BJHS (3.6 ± 0.5 cm) than for controls (1.3 ± 0.2 cm) ($p < 0.01$). Similarly, the Beighton score was greater for BJHS patients (6.6 ± 0.8) than for controls (0.4 ± 0.2) ($p < 0.001$). Hamstring cross sectional area was similar for BJHS (25.1 ± 1.1 cm^2) and controls (26.4 ± 1.6 cm^2).

During the stretch maneuver, the maximal joint angle was greater for BJHS than for the controls ($p < 0.001$), and simi-

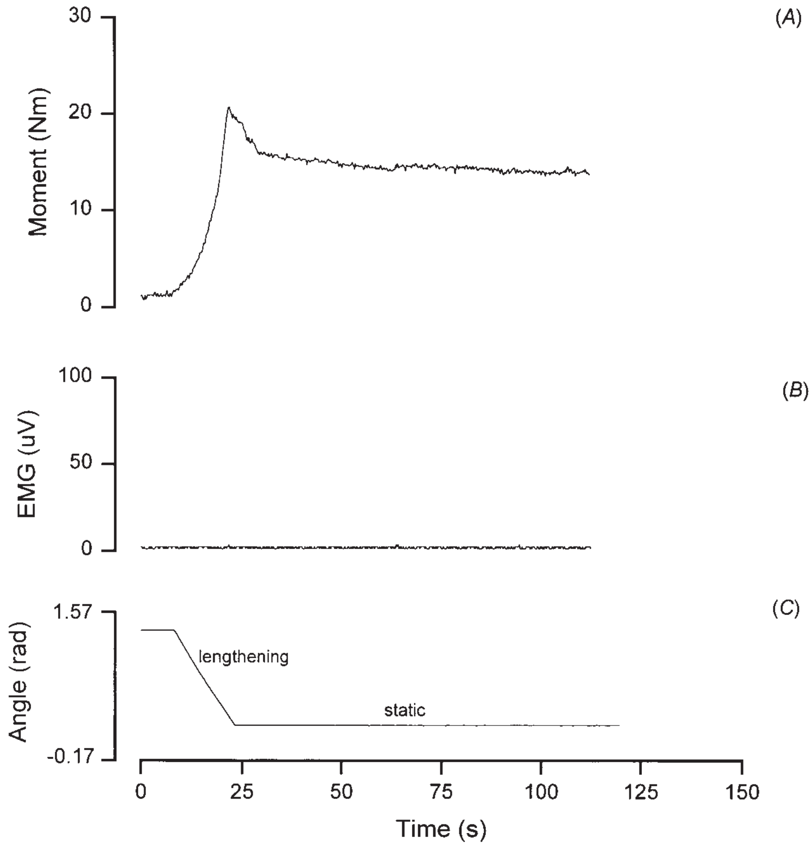

Figure 1. Sampled raw data from a subject during the stretch maneuver. A. The passive moment (gravity corrected) recording with the peak and final moment at the end of the static phase. B. The corresponding EMG amplitude. Note the absence of activity despite the increase in moment in the lengthening phase, and the decline in moment in the static phase (viscoelastic stress relaxation). C. The angle of the stretch maneuver (negative value indicates angles above horizontal) with its lengthening and 90 s static phase.

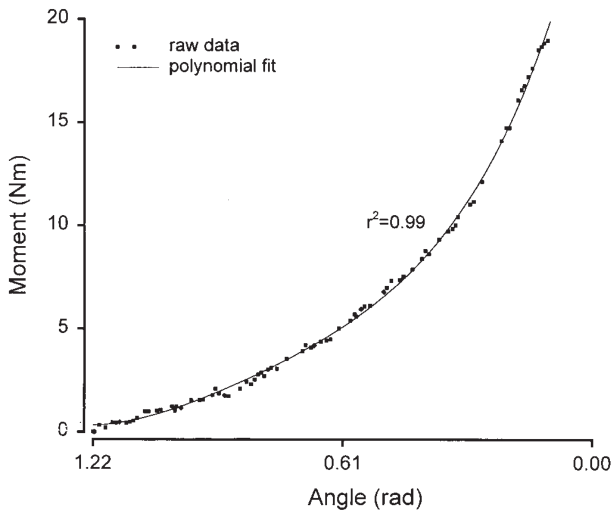

Figure 2. Sample raw data from one subject during the lengthening phase of the stretch. A 4th order polynomial fit was employed. The area under the curve represents the potential energy return from the material.

larly the corresponding peak moment was greater for BJHS than for controls ($p < 0.01$) (Figure 3). For a given common joint angle there was no difference in the passive energy absorption between the groups (Figure 4), but at the maximal joint angle the total energy was greater for BJHS than for controls ($p < 0.001$) (Figure 5). There was no significant difference in viscoelastic stress relaxation between the groups — BJHS 29 ± 3%, controls 27 ± 3%.

DISCUSSION

The main findings of the study were that the viscoelastic stress relaxation and the passive energy absorption for a given common joint angle of the hamstring muscle group were comparable in patients with BJHS and age matched controls. At the same time patients with BJHS had greater forearm skin extensibility and a greater Beighton score than the controls. That is, the passive properties of the hamstring muscle-tendon unit in the 2 groups were comparable, which means BJHS did not have abnormal passive muscle tension compared to healthy subjects. On the other hand, the maximal joint angle for the stretch, the total area-normalized energy, and the corresponding peak moment were greater for BJHS than for controls,

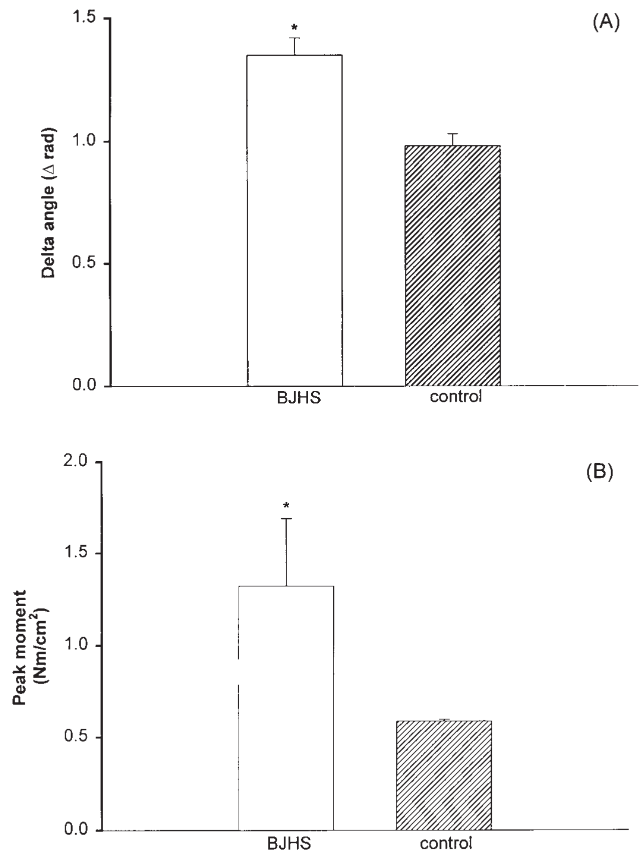

Figure 3. Results from the lengthening phase of the stretch; mean ± SEM. A. The angle of the stretch maneuver was significantly greater for BJHS than for controls. *p < 0.001. B. Maximal peak moment during the stretch maneuver was significantly greater for BJHS than for controls. *p < 0.001.

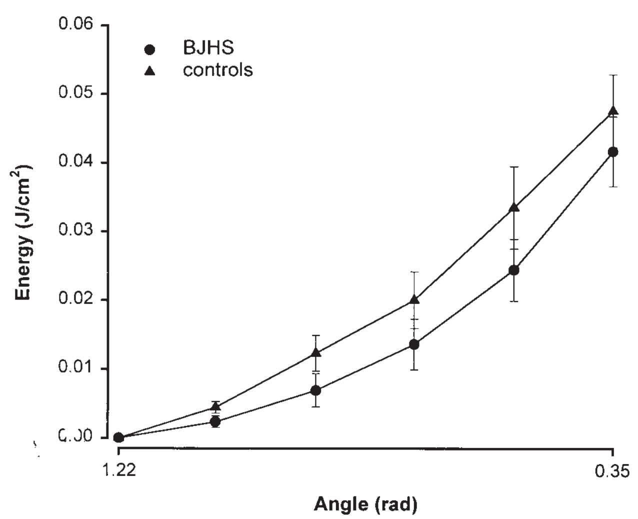

Figure 4. The area-normalized passive energy for a given common angle during the lengthening phase of the stretch; mean ± SEM. There were no significant differences between the groups.

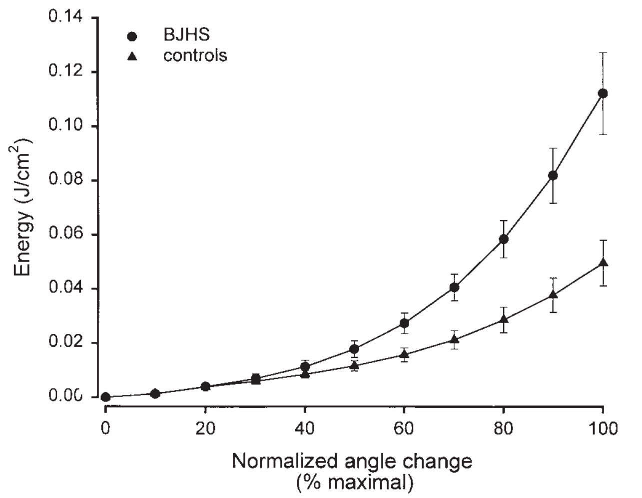

Figure 5. The relative passive energy (normalized to each subject's maximal angle) for the entire range during the lengthening phase of the stretch; mean ± SEM. The maximal energy was significantly greater for BJHS than for controls (p < 0.001). Note that the maximal delta angle for BJHS (1.35 ± 0.07 rad) was greater than for controls (0.98 ± 0.05 rad; Figure 3).

which suggests that BJHS patients displayed a greater subjective tolerance to stretch loading, which may explain the greater flexibility. The subjective tolerance to stretching and passive properties of the muscle-tendon unit have not previously been examined in BJHS.

In addition to joint hypermobility, a common feature of connective tissue disorders, including BJHS, is hyperextensible skin[5,6,23]. Kålund, *et al*[23] investigated the mechanical properties of skin biopsies and demonstrated that for a given tensile load the skin of individuals with Ehlers-Danlos syndrome deformed more than that of age matched controls, which was attributed to reduced mechanical stiffness. Interestingly, the deformation at failure was similar in the patients with Ehlers-Danlos syndrome and controls, which indicates that the skin of Ehlers-Danlos patients is not "hyperextensible," but instead has a reduced strength and stiffness[23]. In our study the clinical forearm skin "extensibility" test was significantly greater in the BJHS subjects (3.6 cm) compared to the controls (1.3 cm), suggesting that the skin of BJHS subjects had altered mechanical properties, which is conceivably the result of an altered collagen synthesis or structure.

The cardinal clinical feature in BJHS is the presence of hyperextensible joints[5,6]. Although never confirmed, this increased musculoskeletal flexibility is commonly believed to be the consequence of modified mechanical properties of the connective tissue in the muscle-tendon unit and joint capsule[8,9]. Interestingly, our study showed that there was no difference in the passive energy absorption of the muscle-tendon unit between the groups for a given common joint angle, although the BJHS subjects achieved a greater maximal angle (Δ1.35 rad) than the controls (Δ0.98 rad). The comparable viscoelastic stress relaxation in the static phase of the stretch emphasizes that the passive properties of the muscle-tendon unit in the 2 groups are similar. The lack of difference in passive properties of the muscle-tendon unit in the 2 groups introduces the notion that the connective tissue of the muscle-tendon unit is unaffected in BJHS.

One of the 2 major diagnostic criteria for BJHS[5,6] is the Beighton score[24]. The average Beighton score in this study was 6.6, which is in agreement with previous reports[4]. However, it should be noted that these tests primarily assess joint capsular involvement, and not necessarily muscle-tendon "looseness." In this study subjects were seated with the trunk and thigh in a position that prohibited complete knee extension. By stretching the hamstring muscles across both the hip and knee joint complete knee extension was avoided, and therefore the muscle-tendon unit was the major contributor to the resistance, or passive energy absorption. Consequently, it is possible for an individual to score high on the Beighton score in the absence of any altered passive properties of the muscle-tendon unit, which is in accord with our findings. Many clinical manifestations of BJHS, including pain and tears of muscle and tendon[5,6], indicate that the muscle-tendon is involved although it has not been measured directly before this study.

As expected, the angle that provoked a maximal stretch sensation was greater for the BJHS subjects than for the controls. However, the greater angle was also accompanied by a larger peak moment (Figure 3) and maximal passive energy in the muscle-tendon unit (Figure 5). The greater peak moment suggests that the subjective tolerance to stretch loading in BJHS subjects differs from the controls. It has previously been shown that subjective tolerance to stretch loading contributes to differences in flexibility[14], and this explains acute and longterm improvements in flexibility[15]. The structures and mechanisms responsible for the subjective tolerance to stretch loading are presently unknown.

It remains unexplained if the many orthopedic problems associated with BJHS are related to the connective tissue of the muscle-tendon unit, joint capsule, articular cartilage, or a combination of them. Interestingly, joint laxity normally decreases with age, while pain appears to worsen in these patients[1,25]. Excessive joint motion, including dislocations, may result in excessive wear of the joint cartilage surface, and thereby contribute to joint arthralgia. The joint capsule and ligaments provide passive constraints to excessive joint motion, while the muscle-tendon unit can contribute with its contractile component to provide joint stability. However, in addition to the contractile component, the muscle-tendon unit has passive properties that can alter joint motion. It has been shown in both animal and human models that resistance training can increase passive muscle-tendon stiffness[26,27]. However, it remains to be established if resistance training, which can increase muscle-tendon stiffness, may also assist in reducing excessive joint motion and the progression of joint pain in BJHS.

While we failed to show that the BJHS significantly affected passive resistance to stretch, it should be kept in mind that the sample size was rather small (n = 9). Consequently, it is appropriate to address the issue of statistical power of the study, or the ability to avoid a type II error. Exactly what represents a clinically relevant difference between the 2 groups is difficult to ascertain. On average the BJHS subjects reached a maximal joint angle that was ~35% greater than that of the controls. Our data yielded a statistical power of ~90% if a 35% lower passive energy was considered to be a clinically meaningful difference.

We examined passive energy absorption of the hamstring muscle-tendon unit in 9 women with BJHS and 9 age and sex matched controls. Forearm skin extensibility and the Beighton score were greater for BJHS than for controls. Also, maximal hamstring stretch angle and the corresponding peak moment were greater for BJHS than for the controls. However, for a given mutual stretch angle the passive energy absorption was the same for both groups, which suggests that passive properties of the muscle-tendon unit are similar. However, the greater joint range of motion and corresponding peak moment in BJHS suggests that BJHS subjects have a greater tolerance to passive moment. Therefore, increased flexibility in BJHS cannot be explained on the basis of altered passive properties.

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