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Abstract

Purpose: Joint hypermobility is a condition in which synovial joints move beyond normal limits. Approximately 10-25% of children and 5-25% of adults experience hypermobility syndrome. One such hypermobility syndrome is an inherited connective tissue disorder known as Ehlers-Danlos Syndrome (EDS). Typically, a score of 4-5 out of 9 on the Beighton scale is indicative of hypermobility in adults. Whereas 6 out of 9 is the criteria for children. No significant correlations were found between the systemic features of EDS and the Beighton score. The purpose of this pilot study was to see if an arthrometer could be used to provide quantitative values of joint laxity to differentiate between individuals diagnosed with Ehlers-Danlos Syndrome versus controls. **Methods:** A Mobil-Aider arthrometer was used to quantify anterior and inferior translation of the glenohumeral joint and anterior translation of the talocrural joint. **Results:** Thirteen control participants without EDS and 14 participants diagnosed by a physician with EDS participated. Significant between-group differences and medium to large effect sizes were found for all 3 motions. **Conclusions:** The Beighton score has known limitations as diagnostic criteria for hypermobility syndrome and EDS. Testing with an arthrometer may contribute objective data on the magnitude of hypermobility, not just dichotomous criteria. This clinical data may contribute to the clinical assessment of connective tissue disorders.

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ABSTRACT

Purpose: Joint hypermobility is a condition in which synovial joints move beyond normal limits. Approximately 10-25% of children and 5-25% of adults experience hypermobility syndrome. One such hypermobility syndrome is an inherited connective tissue disorder known as Ehlers-Danlos Syndrome (EDS). Typically, a score of 4-5 out of 9 on the Beighton scale is indicative of hypermobility in adults. Whereas 6 out of 9 is the criteria for children. No significant correlations were found between the systemic features of EDS and the Beighton score. The purpose of this pilot study was to see if an arthrometer could be used to provide quantitative values of joint laxity to differentiate between individuals diagnosed with Ehlers-Danlos Syndrome versus controls. **Methods:** A Mobil-Aider arthrometer was used to quantify anterior and inferior translation of the glenohumeral joint and anterior translation of the talocrural joint. **Results:** Thirteen control participants without EDS and 14 participants diagnosed by a physician with EDS participated. Significant between-group differences and medium to large effect sizes were found for all 3 motions. **Conclusions:** The Beighton score has known limitations as diagnostic criteria for hypermobility syndrome and EDS. Testing with an arthrometer may contribute objective data on the magnitude of hypermobility, not just dichotomous criteria. This clinical data may contribute to the clinical assessment of connective tissue disorders.

INTRODUCTION

Clinicians frequently assess joint mobility and function. Mobility can span the gamut from immobile to hypermobile.¹ Furthermore, it is important to distinguish the individual who is trained for muscular flexibility from those with generalized articular hypermobility. The value of this differential diagnosis cannot be understated.² Systemic joint hypermobility is a chronic condition and may require lifelong support.²

Joint hypermobility is a condition in which synovial joints move beyond normal limits.³ In children, 10% to 25% experience hypermobility syndrome.^{4,5} Adult hypermobility is reported to range from 5% to 25% in the USA, 25% to 38% in Iraq, and 43% in the Noruba tribe in Nigeria.⁶⁻¹⁰ Cooper and Brems found 76% of surgical patients with multi-directional glenohumeral instability demonstrated generalized joint hypermobility.¹¹ This large percentage is significant and may represent the importance of identifying EDS/hypermobility prior to a surgical procedure.

Joint hypermobility syndrome includes inherited connective tissue disorders such as Ehlers-Danlos Syndrome (EDS).¹² EDS affects many systems of the body and is a complex diagnosis.¹³⁻²² The 2017 International Classification recognizes 13 subtypes of EDS.²³ The Villefranche subtypes include: classical, hypermobility, vascular, kyphoscoliosis, arthrochalasia, and dermatosparaxis.²⁴ The hypermobility type (hEDS) is the most common and represents 80% to 90% of EDS cases.²³⁻²⁵ Individuals with EDS often have poor muscle definition and adopt end-range postures.³ A typical standing posture³ may include flat feet, hyperextended hips and knees, increased lumbar lordosis, and “hip hanging.” Traditional clinical diagnostic criteria have included the Beighton Scale (figure 1) and Beighton Criteria (figure 2). However, the diagnosis of joint hypermobility should also include examination of skin elasticity, scars (thin), stretch marks (adolescent growth spurts), hernia, pelvic floor, varicose veins, Gorland’s sign (tip of the tongue to the nose), and the absence of a frenulum.²³⁻²⁴ Some of these items now appear in the second criterion of the Beighton diagnostic criteria from 2017. Typically, a score of 4 or 5 out of 9 on the Beighton scale is indicative of hypermobility in adults, whereas 6 out of 9 is the criteria for children.²⁶ No significant correlations were found between the systemic features of EDS and the Beighton score.²⁶ Furthermore, the Beighton Score does not differentiate between congenital articular instability versus trained hypermobility. Factors that influence the Beighton Score may include:

1. An individual with EDS may not demonstrate a “positive” score because of muscular guarding/tightening as a protective factor (e.g.: hamstrings in palms to floor test).
2. Individual anatomy may limit people with true connective tissue disorders in instances such as bony end feel (elbow extension or knee hyperextension).
3. People who may have trained for enhanced muscular flexibility (dancers, gymnasts) and do not necessarily have joint instability. Thus, they may score high on this test without the dangers of subluxation or dislocation.
4. The test currently examines a series of joints that are not the most typical of dislocations/subluxations. The Beighton Score does not address the shoulders, hips, or ankles (most problematic lax joints).

Figure 1. Beighton Scale

1. right thumb to radius
2. left thumb to radius
3. right 5th digit hyperextension >90 degrees
4. left 5th digit hyperextension >90 degrees
5. right elbow hyperextension >15 degrees
6. left elbow hyperextension >15 degrees
7. right knee hyperextension >15 degrees
8. left knee hyperextension >15 degrees
9. palms touch the floor with knees straight

Figure 2. Beighton Criteria

<p>Major Criteria:</p> <ul style="list-style-type: none">• Beighton score ≥ 4 out of 9• Arthralgia present in ≥ 4 joints for 3 months <p>Minor Criteria:</p> <ul style="list-style-type: none">• Beighton score ≤ 3 out of 9• Arthralgia present in ≤ 3 joints (or back pain) for ≥ 3 months• Dislocation/Subluxation of ≥ 1 joints, ≥ 1 times• ≥ 3 soft tissue lesions (bursitis, epicondylitis, tenosynovitis)• Marfanoid habitus<ul style="list-style-type: none">○ Wingspan to height ratio > 1.03○ Upper:Lower segment ratio < 0.89○ (+) Steinberg sign• Abnormal skin: hyperextensibility, scarring• Eye signs: eyelids drop, myopia• Varicose veins; hernia, uterine, or rectal prolapse

The purpose of this pilot study was to see if an arthrometer could be used to provide quantitative values of joint laxity to differentiate between individuals diagnosed with Ehlers-Danlos Syndrome versus controls. The arthrometer was used to quantify joint laxity of the shoulders and ankles in individuals with EDS versus a control group.

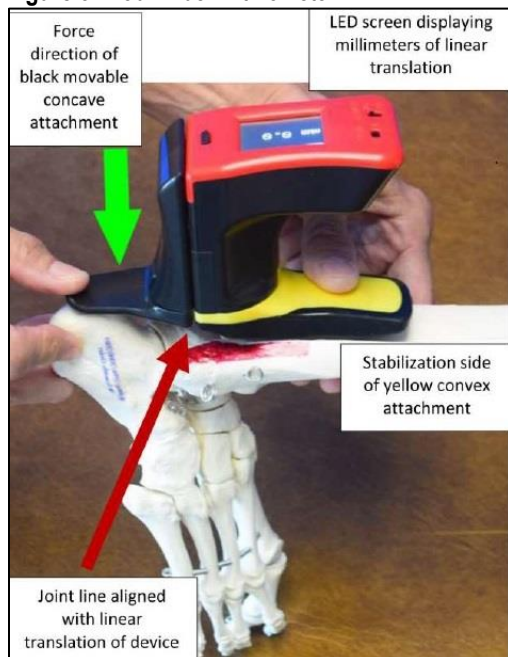
METHODOLOGY

Participants

The consent form, approved by the Institutional Review Board for the Protection of Human Subjects (#87-22) was reviewed and signed by all participants. Each person was screened for the following inclusion criteria. All participants were over 18 years of age. All participants were assessed with the Beighton Scale. All Beighton scores were determined by the researchers (testing researcher was blinded to the Beighton Scale score). Participants in the control group were required to have a “zero” score and no prior injury or surgery to the shoulder or ankle. Participants in the EDS group were diagnosed by a licensed physician. Although they were expected to have a high Beighton score, shoulder or ankle joints with a current injury or prior surgery were not tested. Thus, both shoulders and ankles were tested on some people but not all participants.

Instrumentation

The device used in this study was the Mobil-Aider arthrometer (figure 3). This arthrometer has a stable side (red side with LED screen) and a side that moves linearly (black side without screen) via an internal rollerball mechanism. Each side of the main body of the device accommodates contoured attachments for a variety of joints. In this study ankles and shoulders were tested. For the ankle, the yellow convex attachment contours to the posterior distal tibia (gastroc/soleus/Achilles region) while the black concave attachment conforms to the talus/calcaneal region. Both pieces were locked into position on their respective sides of the device via a dovetail fit and plugger mechanism. The axis of the Mobil-Aider was aligned with the talocrural joint line. The proximal side (yellow) of the Mobil-Aider was stabilized against the posterior tibia. The distal side (black) of the Mobil-Aider was held in contact with the talus/calcaneus. For the shoulder, an inferior translation was performed with the green contoured attachment on the proximal side and the blue attachment was used for anterior translation.

Figure 3. Mobil-Aider Arthrometer**Measurements**

Participants were positioned comfortably for the three testing procedures in this sequence.

- Shoulder inferior translation = supine with arm relaxed at their side, hand on the belly with forearm pronated, and a towel roll under the elbow.
- Shoulder anterior translation = prone with the arm at their side and a small wedge under the ipsilateral clavicle/anterior chest
- Ankle anterior translation = prone with feet over the edge of the table and a small wedge placed under the distal lower leg

The axis of motion of each joint was identified with the passive range of motion performed by the researcher. The Mobil-Aider arthrometer axis was aligned with the joint line. The proximal element of the Mobil-Aider was stabilized against the proximal bone as follows:

- Shoulder inferior translation = stabilize upper thorax/upper chest via proximal aspect of the arthrometer (figure 4)
- Shoulder anterior translation = stabilize scapula via proximal aspect of the arthrometer & wedge inferior to clavicle (figure 5)
- Ankle anterior translation = stabilize tibia (figure 6)



Figure 4. Shoulder inferior translation



Figure 5. Shoulder anterior translation



Figure 6. Ankle anterior translation

The distal segment was mobilized as follows:

- Shoulder inferior translation = apply a distal force through the humeral head (figure 4)
- Shoulder anterior translation = apply an anterior force to the posterior humeral head (figure 5)
- Ankle anterior translation = apply an anterior force through the talus/calcaneus (figure 6)

A few small amplitude test oscillations were performed to confirm proper positioning. Then three movements of each motion (shoulder inferior translation, shoulder anterior translation, ankle anterior translation) were performed with a 30-second rest between tests. Each data point was recorded. Measures were reported in millimeters of linear translation. After the testing of each individual, the surfaces of the Mobil-Aider™ and wedges were cleaned with anti-microbial wipes.

Data Analysis

All data was analyzed using SPSS version 27 (IBM Corp., New York). The average of 3 trials was taken for each motion and then Mann-Whitney U tests were performed to identify between-group differences for all three joint translations measured: anterior and inferior shoulder glide and anterior ankle glide. Effect sizes were calculated using Cohen's *d* formula: $Cohen's\ d = (M_1 - M_2) / S_{pooled}$ where $S_{pooled} = \sqrt{[(s_1^2 + s_2^2) / 2]}$.²⁷ Effect size r_{VI} was then calculated using the formula $r_{VI} = d / \sqrt{(d^2 + 4)}$. Significant between-group differences and medium to large effect sizes were found for all 3 motions (table 1).

Table 1. Between-group differences of joint laxity as tested with an arthrometer

	Control		EDS		Statistical Results	
	Mean	SD	Mean	SD	p-value	Effect size
Anterior shoulder translation	5.45	1.43	10.56	1.74	<.001	.85
Inferior shoulder translation	4.27	1.60	8.51	1.63	<.001	.80
Anterior ankle translation	5.36	1.19	8.07	1.84	<.001	.66

A priori power analysis concluded that 42 total participants would be needed given an assumed effect size of 0.8, the desired power of 0.8, and an alpha level set at 0.05.^{28,29} The post-hoc analysis affirmed that the study was sufficiently powered with 99% power for all data.

RESULTS:

Thirteen control participants without EDS and 14 participants diagnosed with EDS participated in the study. In the control group, 6 participants were male and 7 were female. In the EDS group, 1 participant was male, and 13 were female. The mean age of the control group was 24.1 (\pm 3.4) and of the EDS group was 32.4 (\pm 12.1). In cases where both shoulder or ankle joints met inclusion criteria, these measurements were recorded as a separate case. Thus, for shoulder anterior translation there were 23 in the control and 26 in the EDS group. For shoulder inferior translation there were 21 in the control and 26 in the EDS group. Finally, for the ankle anterior translation, there were 22 in the control and 25 in the EDS group. An independent samples t-test demonstrated a significant between-group difference for age ($p = .026$). The control group was required to have a Beighton score of 0; the EDS group was required to have a Beighton score greater than 6. The EDS group had a mean Beighton score of 8.0 (\pm 1.2). The between-group differences for all joint translations (anterior shoulder, inferior shoulder, anterior ankle) were statistically significant. In fact, the magnitude of the joint translations for the individuals with high Beighton scores were almost double that of those in the control group (Table 1).

DISCUSSION

Joint hypermobility is a topic of interest in the arts, sports, and medical communities.³⁰ However, the lack of awareness of hypermobility syndrome among healthcare providers can be problematic.³¹ Individuals are told their symptoms are "growing pains," "all in your head," or they are "malingerers."³¹ Some individuals have reported they feel their healthcare provider is dismissive or has "given up" on them.³¹ Furthermore, when an individual has hypermobility syndrome, they may be conflicted on whether to participate in sports activities or protect themselves from injury. This can be particularly problematic for parents of children with hypermobility syndrome.

To date, the Beighton scoring system is the most common tool used for the identification of generalized joint hypermobility (GJH). When it was developed in 1973, it was proposed as an epidemiological screening tool, not a clinical tool.³² The Beighton Score is one of the two major components of the Beighton Criteria and is used for the diagnosis of joint hypermobility syndrome and the hypermobility type of EDS.³³ However, despite numerous studies, the cut-offs that differentiate individuals with and without GJH have not been well defined. The range in the literature is from >4 to >8 .^{34,35} When using a Beighton cut-off score of >4 for the entire population, a high false-positive rate of 60% occurred, suggesting an overestimation of prevalence.³⁰ Singh et al (2017) studied 1000 individuals from 3-101 years of age.³² A logistic regression indicated a false-positive rate of 60.0% and a false-

negative rate of 12.4%, with the Beighton scoring system having a sensitivity of 0.8% and a specificity of 99.3% when a cut-off of >4 was used to determine GJH. Based on the Australian cohort for females, the following Beighton scores for GJH were suggested:

- >6 for females & >5 for males aged 3-7 years
- >5 for females & >4 for males aged 8-39 years
- >4 for females & >2 for males aged 40-59 years
- >3 for females aged 60-69 years; >1 for males 60+ years
- >2 for females aged 70+ years

Thus, a single cut-off score does not appear to be sufficient. In addition, the Singh et al³² study did not address ethnic differences. The Beighton system also samples a limited number of joints in a single plane of motion. Commonly lax joints such as shoulders, hips, and ankles are not assessed. The purpose of this study was to pilot the use of additional objective data on joint mobility to potentially contribute to the screening and/or diagnosis of a connective tissue disorder. Objective data is valuable and as technology advances, instruments can provide data to assist clinicians with the quantification of joint laxity. This study used a Mobil-Aider arthrometer to assess multiple joints (ankle in 1 plane & shoulder in 2 planes) and revealed a statistically significant difference between the individuals with and without high Beighton Scores. The mean joint translation of the EDS group was close to double that of the control group (table 1). This magnitude of translation may result in an increased likelihood of joint pain, sprains, subluxations, and dislocations. Early identification with objective criteria could help to identify strategies to reduce these risks.

CONCLUSION

In summary, this pilot study, was intended to see if there were truly objective increases in joint mobility in individuals diagnosed with EDS. Given the recent availability of a joint arthrometer to test joints other than just the knee (KT1000), it will take time to populate the data with normative values across multiple joints. Recent Mobil-Aider arthrometer publications related to knee laxity (+/- 5%; strong correlation with MRI), ankle sprains (difference between grade I & II sprain, $p < 0.004$), shoulder comparisons to electromagnetic devices (ICC=0.83), and wrist inter/intra-rater reliability are steps in that direction.³⁶⁻⁴⁰ Being able to assess joints not addressed by the Beighton Scale may also be helpful. Although this study was adequately powered, additional studies need to be done prior to considering arthrometer values as a diagnostic criterion for connective tissue disorders. Nonetheless, objective data enhances our ability to make clinical decisions and the use of an arthrometer may be able to contribute.

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