Joint hypermobility: The use of a new assessment tool to measure lower limb hypermobility

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ABSTRACT

Objectives

The aim of the study was to compare the use of a new assessment tool for diagnosis of hypermobility in the lower limb to the Beighton score for generalised hypermobility.

Methods

Three groups of children were compared (n = 225) and included a "normal" population of 116 school children, a "possible hypermobile" group of 88 children attending a foot and gait clinic and a "known hypermobile" group of 21 children referred from a paediatrician or rheumatologist. The Beighton score was used to measure generalised hypermobility. The Lower Limb Assessment Score was used to measure hypermobility in the lower limbs.

Results

The Lower Limb Assessment Score was able to distinguish between the three groups of children better than the Beighton score. At a threshold of 5/9 indicating hypermobility, the Beighton score identified hypermobility in 34% of school children; the lower limb score identified hypermobility in 21% of school children after a threshold was identified. There was disagreement between the scores in school children where 26.7% of children appeared to have a positive Beighton score that was not accompanied by a positive lower limb score. In the "known hypermobile" group the Beighton score was positive in only 10% of children when the lower limb score was negative for hypermobility.

Conclusion

In this group of school children, the Beighton score appeared to over-diagnose hypermobility at the threshold of 5/9. Specific thresholds for diagnosis need to be set dependant on the age and ethnic group of the population being studied. The Lower Limb Assessment Score may be a useful score for health professionals specifically interested in lower limb hypermobility.

Key words

Children, hypermobility.

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Introduction

A joint is described as hypermobile if the range of motion is excessive when taking into account the age, sex and ethnic group of the subject (1). Hypermobility is more common in females than males and is known to reduce with age (2). Children therefore, are more flexible than adults but hypermobility can still be present when the joint range of motion is excessive. Hypermobile children suffer from similar joint complaints to adults (3) and the lower limb is affected most often (4,5). In one prospective study, up to 40% of hypermobile children developed symptoms of arthralgia in the course of one year (6).

Diagnosis of hypermobility is often made using the criteria developed by Beighton *et al.* (2). A Beighton score of 5/9 is most frequently used to define hypermobility but this arbitrary cut-off point may make the system insensitive, being inappropriate for different ages and different ethnic groups. The Beighton scale is also heavily weighted to measurement of the upper limb despite the majority of musculoskeletal complaints presenting in the lower limb. With this scoring system, only knee hyperextension is measured in the lower limb and thus subjects presenting with lower limb complaints may fail to be diagnosed if the hypermobility is confined to the lower limb.

Since many of the symptoms related to hypermobility present in the lower limbs, a valid and reliable tool that measures hypermobility in the lower limb would be useful to professionals who diagnose and treat lower limb pathology. This study tested the usefulness of a new assessment scale for lower limb hypermobility. The scale included movements of the joints occurring in several planes of motion rather than in just one direction. The movements included were typical of those used by health professionals such as rheumatologists, physiotherapists and podiatrists, when assessing the lower limb. The score takes around 15 minutes to complete when the examiner is familiar with the set criteria but some initial training may be required if the examiner is not familiar with the positioning used to isolate the joints, such as for knee rotation and midtarsal joint movements.

A comparison of the new scoring system to the Beighton score was made. Through comparison of the two scores, the study aimed to investigate whether the Beighton score was a reasonable measure of lower limb hypermobility and aimed to investigate whether a lower limb scoring system would fail to identify any children defined as hypermobile by the Beighton score. In order to validate the lower limb scoring system, it was compared to the Beighton score as well as with expert clinical opinion as to whether a child was hypermobile. Although most of the scoring systems are best used as a sliding scale of flexibility, the new assessment was tested for validity after a specific cut-off point was set.

Method

Ethical approval was granted by the local ethical committees.

Subjects

Three groups of children were included in the study and were chosen to represent differing levels of hypermobility. The first group consisted of children in primary classes at three London schools and were included to represent "normal". The second group included children attending a paediatric foot and gait clinic at a teaching hospital where a high prevalence of foot conditions related to hypermobility were seen but no child was diagnosed with hypermobility at the time of referral. This group were "possible hypermobile". The third group consisted of children known to be hypermobile, being diagnosed with hypermobility by a rheumatologist or paediatrician. This group were "known hypermobile". The method for assessment for this diagnosis was not known and might vary between referring sources. Usually a full joint examination had been carried out but if a specific scoring system was used, the score was rarely given in the referral.

In the school children, all children attending school on the days of the assessments were included if consent had been given by the parents, unless the child refused to consent on the day or met any of the exclusion criteria. Consent forms were sent to all parents of children attending the primary schools and requested basic demographic information including child's age, sex and ethnic group. Incomplete forms were supplemented from school records. The children were assessed in a quiet classroom environment within their schools. For the children attending the paediatrics clinic, all children attending for routine appointments over a 6 month period were assessed if informed consent was given by the parent and child and the exclusion criteria were not met. Consent was obtained from all children attending the clinic at the time of their clinical appointment and demographic information was collected at that time.

Children were excluded from the study when:

- A diagnosis of joint disease (i.e. Juvenile Idiopathic Arthritis) or connective tissue disease existed that might restrict normal joint motion.
- A current or previous orthopaedic condition (other than simple fracture) was present influencing normal movement (ie. talipes, tarsal coalition).
- The child had a diagnosis of neurological or neurodevelopmental disorders.
- The child was aged less that 5 years old or older than 16 years old.

Hypermobility assessment

All children were assessed using the Lower Limb Assessment Score providing a score to a maximum of 12 marks for each limb. The Beighton score was taken for all children providing a single score to a maximum 9 (see Appendix 1). Each child was also graded according to the clinical opinion of the assessor using three gradings (hypermobile, borderline hypermobility, normal) by undertaking a full lower limb examination prior to the application of any hypermobility score.

Two examiners undertook the assessments after inter-observer repeatability of the Lower Limb Assessment Score was tested using reliability analysis in a pilot study of 22 children (Intraclass correlation coefficient of 0.84 (95% CI = 0.62 to 0.93; F (1, 21) = 6.08, p = 0.001)).

Statistical analysis

The distributions of the data were initially examined for normality and parametric tests or non-parametric tests carried out based on the distributions found. P values less than 0.05 were considered statistically significant.

Results

The "normal" group included 116 children (66♀: 50♂) with a mean age of 7 years (SD 1.9 years). The "possible hypermobile" group included 88 children (46♀: 42♂) with a mean age of 9.89 years (SD 3.39 years). The "known hypermobile" group included 21 children (13♀: 8♂) of mean age 9.18 years (SD 3.55 years).

Association between the Beighton and LLAS scores with age

The clinic children were slightly older than the school children therefore associations with age for the Beighton score and the LLAS score were tested initially. Pearson correlation was used to test for an association. No association was found in any of the groups for either the Beighton with age ("normals": r= 0.12; "possible hypermobile": r = 0.36; "known hypermobile": r = 0.2) or the LLAS with age ("normals": r = 0.21; "possible hypermobile": r = 0.46; "known hypermobile": r = 0.19).

Comparison of left to right sides for the Lower Limb Assessment Score

A paired t-test was used to test the null hypothesis of no significant difference occurring between left and right sides scores (n = 225) for the LLAS. The null hypothesis was accepted: no significant difference was found (p = 0.74). The LLAS was taken as a score out of 12 rather than a total score of 24 for the two limbs for further analysis.

Using the Beighton score to measure hypermobility in children

The numbers of children found to be hypermobile using the Beighton criteria were compared in the three groups. Hypermobility was defined as a score of 5/9 or greater (Beighton positive). Within the school children ("normals"), 39 children (34%, 95%CI = 27 to 41%) were found to be hypermobile using this criteria for hypermobility. In the group "possible hypermobile", 31 children (35%, 95%CI = 25 to 45%) were found to be hypermobile whilst in the group referred from a rheumatologist/paediatrician ("known hypermobile"), 11 children (52%, 95%CI = 31 to 74%) were Beighton positive.

The percentage of children at each level of the Beighton score was used in order to see if the distribution of the score across a group could differentiate between the three groups of children. Figure 1 suggested that the Beighton score was not able to clearly differentiate between the three groups of children with the "normals" and "possible hypermobiles" having a similar distribution. At the higher scores, the percentage of children increased in the "known hypermobile" group and decreased in the other two groups showing some distinction between the groups.

A Kruskal-Wallis test was used to test the null hypothesis of no significant difference being detectable between the three groups. The null hypothesis was rejected: the probability value obtained suggested that the difference between the groups was just on the level of significance ($\chi^2 = 5.87$, df = 2, p = 0.053).

Considering if the Lower Limb Assessment Score is a reasonable measure of hypermobility in children

The Lower Limb Assessment Score was investigated with regard to differentiating between the three groups of children. Observation of figure 2 suggested that the LLAS was able to distinguish more clearly between the groups. A Kruskal-Wallis test was used to test the null hypothesis of no significant difference occurring between the three groups of children. The null hypothesis was rejected: a significant difference was seen between the groups ($\chi^2 = 21.52$, df = 2, p < 0.001).

Defining a cut-off score to represent hypermobility with the LLAS

In order to consider the number of children that would be diagnosed with hypermobility using the Lower Limb Assessment Score, a threshold to define hypermobility had to be set. All chil-

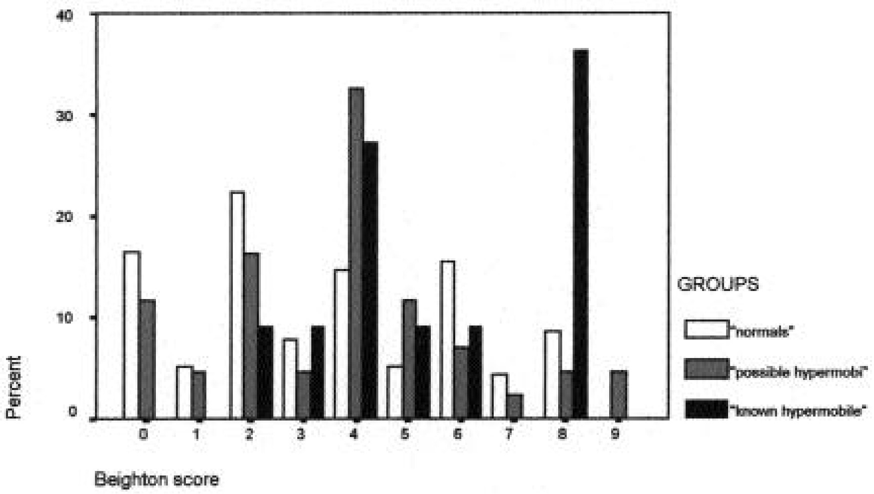

Fig. 1. Bar chart showing Beighton scores in three groups of children.

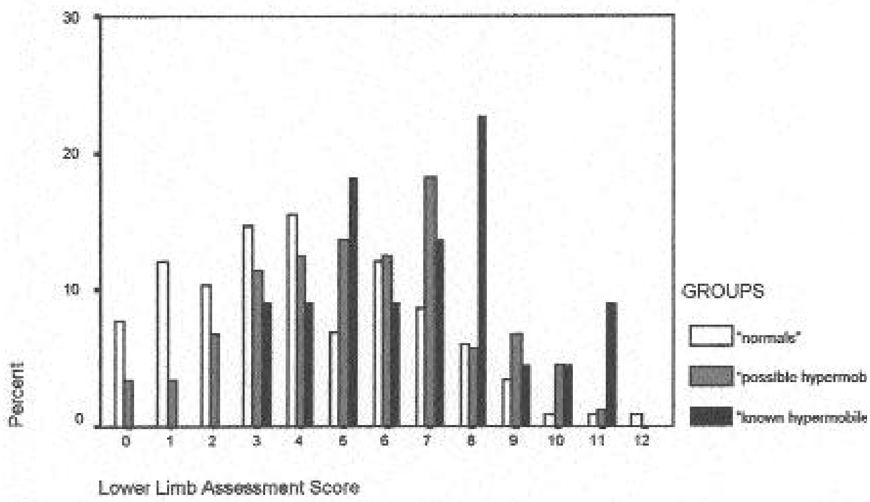

Fig. 2. Bar chart showing the Lower Limb Assessment Score for the three groups.

dren were assessed for the clinical opinion of the examiner as to whether they were hypermobile, borderline hypermobility or normal. The Lower Limb Assessment Score within each group was compared.

Figure 3 expresses these mean values graphically, with the thick bar representing the median value and the box defining the 25th and 75th percentile values. The bar ends represent the smallest and largest values that were not outliers.

A cut-off score of 8/12 appeared to give a score than would differentiate between hypermobile children and normal children, with minimal overlap with the borderline cases. Sensitivity analysis was undertaken to determine the numbers of true positive and false positive cases for each level of the score. The sensitivity and specificity for each level of the Lower Limb Assessment Score was tested using ROC analysis. For this analysis, the clinical opinion was used as the gold-standard for comparison. Children who were borderline cases were reclassified as normal for this analysis based upon the decision that if the clinician was undecided as to the diagnosis, the child was probably not hypermobile. Figure 4 shows the ROC curve and Table I gives the specificity and sensitivity levels.

The ROC curve (Fig. 4) and Table I shows how at the cut-off threshold of LLAS = 7, the sensitivity is good at 0.943 (94%) and the false positive rate is 0.075 (7.5%). At the threshold of LLAS = 8, the sensitivity has decreased to 0.704 (70.4%) but the false positive rate is extremely good at 0.004 (0.4%). The ROC curve shows the trade-off between sensitivity and specificity with the curve closest to the left-hand and top border being the most accurate. The point on the ROC curve that is closest to both axes expresses the most useful score in terms of sensitivity and specificity. On this curve, this point is the score of 7/12 for the LLAS.

The positive predictive values and negative predictive values were then considered in each group of children when a cut-off point of 7/12 was used to denote hypermobility. The positive predictive value tests the probability that the subject has hypermobility, when restricted to those subjects who do have hypermobility. Table II shows the values for each group.

With the cut-off score being identified, the percentage of children in each group found to be hypermobile could be identified. The percentages of children in each group when a cut-off point of 7/12 was applied were: "normals" 21% (95%CI=11 to25), "possible hypermobiles" 36% (95%CI= 26to 46) and "known hypermobile" 52% (95% CI=31to74). The percentages of children in each group when a cut-off point of 8/12 was applied were: "normals" 16% (95%CI=10to23), "possible hypermobiles" 38% (95%CI= 27to 48) and "known hypermobile" 41% (95% CI=21to64).

Comparing the Beighton score and the Lower Limb Assessment Score - can the Beighton score diagnose lower limb hypermobility? Does the LLAS fail to diagnose children with generalised hypermobility ?

The level of agreement between the two scores was sought using the Beighton score threshold for hypermobility as 5/9 and the LLAS threshold as 7/12. In the school children ("normals"), there was agreement between scoring systems in 69% of cases. In the remaining cases were there was disagreement, in 31 cases (26.7%), the Beighton score suggested the child was hypermobile but the LLAS suggested that there was no hypermobility present in the lower limbs. In 5 cases (4.3%), the LLAS identified hypermobility but this was not accompanied by a Beighton score of greater than 5/9.

In the group "known hypermobile", there was agreement between the LLAS and the Beighton score in 80% of cases. In 2 children (10%) the Beighton score indicated hypermobility was present when the LLAS found no lower limb hypermobility. In 2 children (10%) the LLAS identified lower limb hypermobility but the Beighton score did not indicate generalised hypermobility.

The association between LLAS and the Beighton score was tested with Pearson correlation. A weak relationship was seen between the scores of the school children (r = 0.43) but a stronger relationship was seen for the "possible hypermobile" (r=0.79) and the "known hypermobile" groups (r = 0.79).

Discussion

The study found some differences between the scores that might be expected given that the scoring systems were measuring different aspects of hypermobility, but also placed some doubt on the use of the Beighton score in population studies. A difference in the levels of hypermobility was expected in the three populations. The "known hypermobiles" were expected to have the highest prevalence of hypermobility given that a diagnosis had been made prior to inclusion into the study, by a paediatrician or rheumatologist. The "possible hypermobiles" were expected to show a higher prevalence of hypermobility than the "normals" due to the numbers of cases of hypermobility seen in the clinics with foot or gait problems. The LLAS was clearly able to differentiate between these populations but the Beighton score could not differentiate between the groups so well. This may have been for two reasons. Firstly, two of the groups were seen in a foot and gait clinic so had problems presenting in the lower limbs – by measuring lower limb joints, the range of lower limb joint flexibility was going to be shown using the LLAS. By only measuring knee hypermobility, it was perhaps unfair to expect the Beighton score to distinguish between the particular groups. Secondly, the school children that were measured came from a predominantly Asian background whilst the children presenting to clinic were predominantly Caucasian. Asians are known to be more hypermobile than Caucasians (7) and thus the increased prevalence of hypermobility in these school children reduced the distinction between the "normals" and the "possible hypermobiles", with only the more extreme cases in the "known hypermobile" group remaining distinct.

The Beighton score appeared to overdiagnose hypermobility in the "normal" population in this study, demonstrated by the high prevalence of hypermobility identified (34%) when other studies have suggested that a figure of 15% to be more acceptable (8). Using a higher threshold or identifying an appropriate threshold through initial studies might reduce the prevalence seen. This was seen with the LLAS where

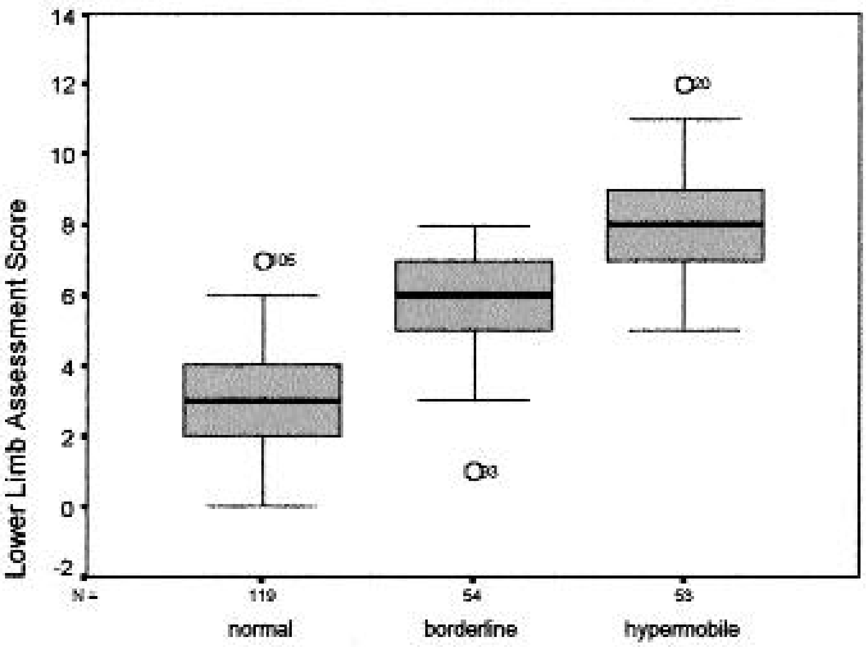

Fig. 3. Differences between the assessment scores for each clinical diagnosis (left side).

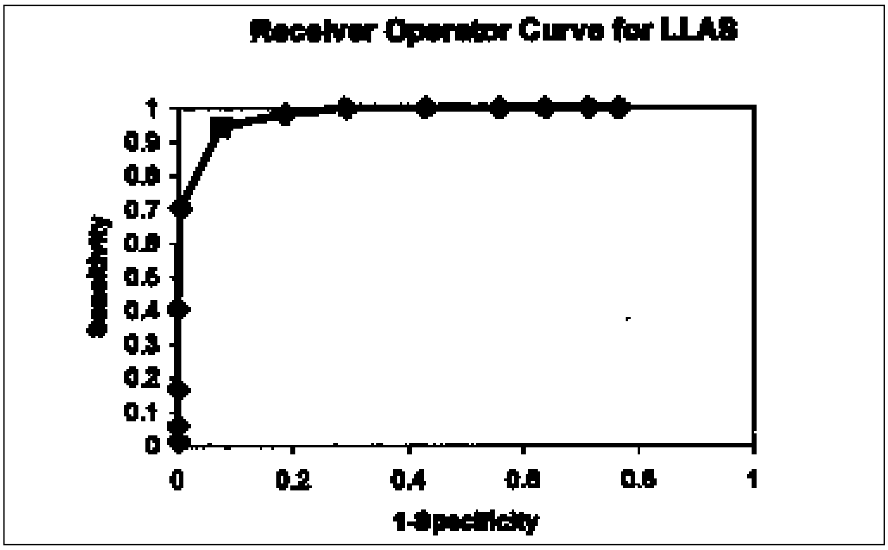

Fig. 4. ROC curve for the Lower Limb Assessment Score.

Table I. Sensitivity (true positive) and specificity (false positive) for diagnosing hypermobility with the LLAS.

Lower Limb Assessment Score		
Cut-off point	Sensitivity	1-specificity
0	1.000	0.765
1	1.000	0.712
2	1.000	0.637
3	1.000	0.558
4	1.000	0.429
5	1.000	0.292
6	0.981	0.186
7	0.943	0.075
8	0.704	0.004
9	0.407	0.000
10	0.169	0.000
11	0.058	0.000
12	0.014	0.000

Table II. Positive predictive values (PPV) and negative predictive values (NPV) for the three groups of children, taken at hypermobility = LLAS of 7/12 or greater.

	PPV	NPV
"Normals"	58.3%	89.1%
"Possible hypermobile"	84.4%	91.5%
"Known hypermobile"	83.3%	83.3%

sensitivity analysis allowed an appropriate threshold to be identified and the prevalence in the "normal" population was at a more acceptable level (21% using a threshold of 7/12 or 16% using a threshold of 8/12). Also suggestive that the Beighton score was over-diagnosing hypermobility at the 5/9 threshold was the level of disagreement between the Beighton and the LLAS in the "normal" population.

It was found that 26.7% of the school children ("normals") were positive for the Beighton score but negative for the LLAS. This would suggest that these children had upper limb/generalised hypermobility but no lower limb hypermobility. In the "known hypermobile" group there was 80% agreement between the scores so that most children with upper limb hypermobility also had lower limb hypermobility. It may be that the school children demonstrate a sub-population who have only upper limb hypermobility but it is more likely that the five tests used in the Beighton score were not sensitive enough to identify hypermobility, being easy to perform by many children. By only including one joint in the lower limb (knee hyperextension) the Beighton score also appeared to miss the diagnosis of hypermobility a small number of children (4.3%) who had lower limb hypermobility.

In life, flexibility is not an "all or none" state but there are degrees of flexibility and it is not possible to define one point when a person becomes hypermobile. However, in population or research studies it is useful to have a threshold for definition. If the LLAS was being used in a research project where and it was essential to exclude all normal children, then ROC analysis should be applied to identify an appropriate threshold for that population initially. Sensitivity and specificity can vary when populations are dramatically different and so it may be appropriate to repeat the analysis for older age groups or different ethnic groups.

Using the cut-off point of 7/12 for the LLAS in this study appeared to give more realistic values for the prevalence of hypermobility in the "normal" group (school children) of 21% but the prevalence of hypermobility in the "known hypermobile" group was low at 52% of the group compared with the Beighton score that found 60% of the group as hypermobile. For both these scoring systems, the prevalence of hypermobility in the "known hypermobile" group would be expected to be higher. It was unclear in the children being referred, how the diagnosis of hypermobility was made. It may have been based on the Beighton score and thus given the findings in the school group where the Beighton score was found to over-diagnose hypermobility, the LLAS would be expected to be lower. If this was the situation, the prevalence with the Beighton score should have been closer to 100%. The lower figure may reflect examiner bias where more strict criteria were applied when measuring the score for the study compared with a clinical situation. Alternatively, the referring practitioner may only have found hypermobility at a particular joint – for example, the symptomatic joint – and given the diagnosis based upon that joint and not applied any scoring system during their assessment.

In conclusion, this study introduced a new scoring system for the diagnosis of hypermobility in the lower limb joints. The scoring system was shown to have benefits over the traditionally used Beighton score and although best used as a sliding scale of flexibility, a cut-off point to diagnose hypermobility in the lower limb was set. The Lower Limb Assessment Score would be useful for prospective studies, when excluding/ including hypermobile participants. It would be a useful clinical tool for health professionals to aid in the diagnosis of lower limb conditions that may be related to joint hypermobility.

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APPENDIX 1

Lower Limb Assessment Score	LEFT		RIGHT	
HIP FLEXION – The patient lies supine; the examiner flexes one hip fully; the other leg must stay fully extended on the couch. Does the mid-anterior area of the thigh drop easily onto the stomach/chest with a loose feel to the movement, using a minimum to moderate application of force?	YES	NO	YES	NO
HIP ABDUCTION – The patient lies supine, with hip and knees flexed; the knees are dropped outwards and down to the couch, the soles of the feet remain together. With the examiner's hand against the lateral femoral condyle, can the knees come down to the couch sufficiently to let the back of the examiners hand touch the couch? - minimal application of force required.	YES	NO	YES	NO
KNEE HYPEREXTENSION – The patient lies supine; the knees are relaxed and straight; With minimal force, keeping the femoral condyles on the couch, can the heel be lifted at least 3cm off the couch (greater than 2 finger widths)?	YES	NO	YES	NO
KNEE ANTERIOR DRAW TEST – The patient is supine; the hips and knees (90°) are flexed; the examiner gently sits of the foot to stabilise it; moderate pressure is placed against the femoral condyles as the tibia is pulled forwards. Is there a definite, obvious forward movement of the tibia against the femur? Palpable "clunking" of the joint surfaces moving against each is indicative of a positive draw sign.	YES	NO	YES	NO
KNEE ROTATION – The patient lies supine; the examiner flexes the hip and knee to 90° and palpates the tibial tubercle; holding the malleoli and ankle firmly, the tibia is rotated medially and laterally on the femur. Normal movement is 1cm medially and laterally. Does the tubercle move easily beyond 1cm in any direction or greater than 2cm overall? With increased internal movement the head of the fibula/lateral condyle of the tibia may also be seen to move.	YES	NO	YES	NO
ANKLE JOINTDORSIFLEXION – The patient lies supine; the knee is flexed to 45°; with moderate to strong force the ankle is dorsiflexed. Does the ankle flex more than 15 degrees? Along with the increased movement there may be bulging of the skin and subcutaneous fat anterior to the ankle.	YES	NO	YES	NO
ANKLE ANTERIOR DRAW TEST – The patient lies supine; the knee is flexed to 45°; the examiner grasps the heel along the plantar and posterior surfaces with one hand and applied a stabilising force against the anterior of the tibia with the other hand. Using a strong anterior force, can the calcaneum and talus be brought forwards on the tibia? Any forward movement felt is a positive result.	YES	NO	YES	NO
SUBTALAR JOINTINVERSION – The patient is supine with their feet over the end of the couch; the examiner holds the posterior surface of the heel and moves the heel into inversion without moving the leg. Is excessive inversion of the subtalar joint seen using minimal force? The sole of the foot or visualisation of the neck of the talus should show movement of 45° inwards, the lateral head of the talus will be very prominent.	YES	NO	YES	NO
MIDTARSAL JOINT INVERSION – The patient is supine with their feet over the end of the couch; the midtarsal joint is isolated from the subtalar joint; the forefoot is grasped from lateral to medial along the metatarsals; only minimal - moderate force is applied to invert the midtarsal joint. Does the midtarsal joint invert beyond 45° so that the plantar surface of the metatarsal heads can be brought inwards by 45 degrees?	YES	NO	YES	NO
MIDTARSAL JOINT AB/ADDUCTION AND DORSI/PLANTARFLEXION – The patient is supine with their feet over the end of the couch; the examiner grasps and stabilises the rearfoot; the forefoot is moved in the direction of ab/adduction and dorsi/plantarflexion. Normal movement should be 1cm in each direction. With minimal force, does the forefoot move easily, almost "wobbling", in an increased amount? Excessive movement in either of the two planes is a positive result.	YES	NO	YES	NO
METATARSOPHALANGEAL MOVEMENT – The patient is supine with their feet over the end of the couch; the hallux is dorsiflexed using minimal – moderate force. Does the hallux dorsiflex easily beyond 90° relative to the metatarsal?	YES	NO	YES	NO

APPENDIX 1

LowerLimb Assessment Score	LEFT		RIGHT	
EXCESSIVE SUBTALAR JOINTPRONATION – The patient is to march on the spot and stop on command; the patient is asked to invert their foot and hold the position close to subtalar joint neutral; the patient is then asked to relax their foot; the movement is observed. Does the arch lower and flatten fully, excessively and easily, with the talus bulging medially? The pronation noted should be at the end of range of the subtalar joint motion so that no further pronation is possible	YES	NO	YES	NO
To score, each limb is calculated separately giving a left score and right score. Each YES is given one mark. Atotal of score of 12 marks is available.	TOTAL:			

Beighton Score [taken from Beighton *et al*. (2) and Hudson *et al*. (10)].

PASSIVE DORSIFLEXION OF THE 5TH FINGER M.C.P JOINT – With the palm placed on a flat surface, the 5th finger is raised so that the m.c.p joint is dorsiflexed to resistance using miminal force. An angle of 90 degrees or greater is a positive result.	YES	NO	YES	NO
PASSIVE APPOSITION OF THE THUMB TO THE FLEXOR ASPECT OF THE FOREARM – With the wrist flexed, the thumb is passively moved towards the forearm. The thumb contacting the forearm using minimal force is a positive result.	YES	NO	YES	NO
HYPEREXTENSION OF THE ELBOW – With the subject sitting or standing and the upper arm supported, the forearm is aligned with the upper arm with the elbow in a neutral position. The forearm is then allowed to drop and gentle pressure placed on the forearm to hyperextend the elbow. Extension greater than 10 degrees is a positive result.	YES	NO	YES	NO
HYPEREXTENSION OF THE KNEE – The subject lies supine and the knees relaxed and straight. With minimal force, keeping the femoral condyles on the couch, can the heel be lifted at least 3cm off the couch – 10 degrees hyperextension?	YES	NO	YES	NO
FORWARD FLEXION OF THE TRUNK – With the subject standing, keeping the knees straight, the subject is asked to bend forwards and touch the floor. Placing the palms of the hands on the floor is a positive result.	YES	NO	YES	NO
	One point is given for each "yes", to give a total score out of 9.			