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The hypermobility spectrum in rugby union players, netballers and dancers: implications for injury and performance

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Abstract

Objectives: Hypermobility has been associated with injury and performance and a new hypermobility framework has been introduced. This study aimed to report the prevalence of localised joint hypermobility, generalised joint hypermobility (GJH), peripheral joint hypermobility and hypermobility Ehlers-Danlos type in female rugby players, male rugby players, female netball players, female dancers, male and female controls.

Methods: This study determined joint hypermobility via the Beighton score and the associated criteria of the hypermobility spectrum in 378 participants.

Results: Localised joint hypermobility ranged from 61.11% (netballers), 57.33% (female rugby), 48.15% (male controls), 46.30% (male rugby), 38.33% (female controls) to 28.57% (female dancers). Significant differences existed for Beighton scores ($p < 0.001$) between female dancers and all other cohorts, female rugby and male controls ($p = 0.005$), male rugby and netball ($p = 0.001$), netball and male controls ($p = 0.001$) and female controls and male controls

($p=0.021$). Prevalence of GJH ranged from 69.84% (female dancers), 25% (netball), 21.67% (female controls), 18.67% (female rugby), 3.70% (male rugby) to 1.85% (male controls). In participants with GJH, dancers had the highest prevalence of pain and dislocation/subluxation. Significant differences existed between dancers and all other groups for hypermobility Ehlers-Danlos type criteria ($p<0.001$). Five participants met the criteria for diagnosis of hypermobility Ehlers-Danlos type. Male rugby players had the highest prevalence of peripheral joint hypermobility (29.63%).

Conclusion: Significant findings between dance and other cohorts may highlight a potential performance adaptation. Significant findings between control groups for the Beighton score may indicate a gender effect. There is a need to consider these factors in relation to performance and injury.

Keywords: Beighton score, general joint hypermobility, hypermobility Ehlers-Danlos type, peripheral joint hypermobility, female dancers

Introduction

Joint hypermobility (JH) is the capability of a joint to move passively and/or actively beyond normal limits along physiological axes¹ and is a descriptive term rather than diagnostic.¹ JH is determined via the Beighton and Horan Joint Mobility Index² which provides a Beighton score (BS). The BS assesses five joint movements that provide a maximum score of 9 with scores of ≥ 4 classified as hypermobile.³ However, the literature lacks clarity with values of 4, 5 and 6 defined as hypermobile.⁴ Recently a new spectrum of JH related disorders has been proposed that cluster the phenotypes presenting JH plus one or more of its secondary musculoskeletal manifestations (dislocations, subluxations, soft tissue injuries, chronic pain, disturbed proprioception and bone mass changes) that do not fulfil the criteria for hypermobility Ehlers-Danlos type (hEDS).¹ These spectrums are: localised JH (JH at fewer than 5 types of joints); generalised joint hypermobility (GJH) spectrum disorder (JH in 5 or more joints); peripheral JH disorder (present in hands or feet only) and historical JH disorder (present in adults whom have lost GJH via the aging process and associated range of motion (ROM) reductions). Prior to this framework¹, joint hypermobility syndrome (JHS) was advocated to describe a connective tissue disorder associated with hypermobility in which musculoskeletal complaints are present in the absence of systematic rheumatological disease.⁵ JHS was diagnosed by the Revised Brighton criteria (1998) consisting of 2 major and 8 minor criteria⁶ characterised by

the presence of symptoms including arthralgia, dislocation, subluxation, spinal conditions, soft tissue rheumatism, marfanoid habitus, abnormal skin, eye signs, varicose veins and hernia which may have a detrimental impact on life quality and athletic performance. However as the presence of JH and related musculoskeletal symptoms are not a syndrome use of the spectrum is recommended.¹

JH commonly occurs in hereditary connective tissue diseases that affect connective tissue matrix proteins and includes Ehlers Danlos Syndrome with particular prominence in Type III, Type I and IV of Osteogenesis Imperfecta and Marfan syndrome⁷ which are associated with morphological features and increased joint dislocation, bone fragility and skin hyperextensibility.⁸ Previously some experts considered that JHS and hEDS were the same condition.⁹ Following review, Ehlers-Danlos syndromes are defined as heterogeneous heritable connective tissue disorders associated with JH, skin extensibility and tissue fragility which have recently been classified into 13 subtypes.¹⁰ Previously the Villefranche Nosology¹¹ was used to provide clinical diagnostic of Ehlers-Danlos syndromes which defined six subtypes with associated major and minor criteria with the assumption these subtypes were due to altered fibrillar collagen gene structure or changes in the genes that encoded collagen modifiers. The lack of a genetic defect in Ehlers-Danlos syndromes¹⁰ resulted in a clinical classification utilising previous descriptive names that highlights the characteristic manifestations of the phenotype and the development of major and minor criteria for each subtype.

JH and JHS have increased prevalence in Asians and Africans in comparison to white Caucasians¹² and is more prevalent in females^{13,14} and within sporting populations than the general population. JH has been associated with an increased risk of injury in rugby¹⁵, netball¹⁶ and dance^{17,18} and a meta-analysis demonstrated increased injury risk at the knee but not the ankle in contact sports in JH individuals.¹⁹ In rugby, JH may be a risk factor for injury due to the increased physical contact associated with tackling¹⁵ while a trend towards impaired movement control was reported in netballers with GJH.²⁰ McCormack et al²¹ reviewed the incidence of JHS at the Royal Ballet and concluded that previously injured hypermobile dancers were less likely to progress into the profession and a 5 year follow up of these dancers suggested they were prone to injuries and had more time absent from dancing due to injury.²² Klemp et al¹⁷ reported an injury prevalence of 33.3% in JHS dancers which was significantly greater than non-JHS dancers. Proprioception and neuromuscular control is impaired in hypermobile individuals with Ehlers-Danlos syndrome²³ and the relationship between reduced

proprioception and muscle strength may have a detrimental effect on physical activity levels.¹ Recent research has advocated that the Beighton score may potentially predict arthralgia in female rugby players and female controls and dislocation and subluxation in male rugby players and controls.²⁴ For JH, specific physiotherapy treatment strategies including strapping or bracing may increase joint stability and reduce arthralgias, joint subluxations/dislocations and sprains.²⁵ In sports and activities that require a high degree of flexibility such as dance, hyperflexibility might be beneficial for performance.²⁶ Aesthetic demands may influence the selection of hypermobile dancers for dance schools and within ballet, JH may be an asset²⁷ and its identification can allow sport selection which maximises physical attributes and minimises injury risk.

An enhanced understanding of hypermobility within the new hypermobility spectrum¹ may aid injury prevention and performance management and allow appropriate interventions via strength and conditioning programmes and training load monitoring. The primary aim of the study was to report the prevalence of localised JH and GJH in female rugby players, male rugby players, female netball players, female dancers, male and female controls. The secondary aim was to report the specific secondary musculoskeletal manifestations of GJH and peripheral JH and hEDS prevalence within these cohorts.

Materials and methods

Participants

Three hundred and seventy-eight participants volunteered to participate in this study and their demographics are outlined in table 1. Sport and dance groups were standardised for weekly participation levels of 8 hours per week of training/matches and rehearsals. Controls were recruited by asking for volunteers via a poster campaign within the university. All participants were 18 years of age or older and were excluded from the study if they had suffered an injury in the previous 30 days²⁸ which prevented training, match or dance class participation. Participants completed a medical screening questionnaire prior to participating in the study and additional exclusion criteria included heart disease, pregnancy, rheumatoid arthritis and scoliosis. Informed consent was obtained from all individual participants included in the study. Participation was voluntary and participants were provided with information sheets and completed informed consent forms prior to participation. All procedures performed involving human participants were in accordance with the ethical standards of the institutional research committee and with the 1975 Helsinki declaration as revised in 1983.

Table 1 participant demographics

Group	Age (years)	Height (cm)	Mass (kg)	Ethnicity
FR (n = 75)	20.29 ± 1.76	165 ± 8.63	74.40 ± 17.67	74 white Caucasian 1 black Caribbean
MR (n = 54)	21.24 ± 2.87	180 ± 7.27	89.46 ± 16.49	51 white Caucasian 3 black Caribbean
NP (n = 72)	20.79 ± 1.54	169.49 ± 7.61	66.47 ± 10.91	71 white Caucasian 1 Asian
FD (n = 63)	21.01 ± 1.19	161.83 ± 7.20	59.19 ± 5.22	61 white Caucasian 1 Hispanic 1 Asian
MC (n = 54)	21.01 ± 1.23	175.56 ± 7.92	76.95 ± 9.17	52 white Caucasian 2 black Caribbean
FC (n = 60)	20.61 ± 1.18	163.86 ± 7.83	64.12 ± 9.51	59 white Caucasian 1 black African

Procedures

All testing was conducted indoors under the supervision of the same researcher and prior to testing the participants' height (cm) was measured using a stadiometer (Leicester Height Measure, Child Growth Foundation, Leicester, UK) and body mass (kg) were recorded using digital scales (Salter 9028, Kent, UK). The participants date of birth and ethnicity was recorded and participation in other sports and dance was determined prior to testing to ensure that participants did not cross participate and no individuals were found to cross participate. Testing was conducted prior to training or dance classes to prevent any potential effects of exercise on JH and participants did not participate in exercise for at least 12 hours prior to testing due the potential effects of warm up on joint ROM.²⁹

JH screening

The BS² was used to measure localised JH, GJH, peripheral JH and hEDS. The BS has an Intraclass Correlation Coefficient (ICC) of 0.91 and a kappa score of 0.74.³⁰ The researcher who was a physiotherapist with 16 years of experience in BS classification performed all measurements by measuring ROM of the 5th metacarpophalangeal joints (1 point each joint),

thumbs (1 point each joint), elbows (1 point each joint), knees (1 point each joint) and lumbar spine (1 point) providing a maximum score of 9. A goniometer (Vivomed, UK) was used to measure all joints except the lumbar spine for which JH was classified as yes/no based on the participants ability to put the palms of their hands flat on the floor. All tests were performed as described by Juul-Kristensen et al.³⁰ Intra-rater reliability was calculated using an ICC (3,1)³¹ by the researcher measuring JH using the BS of 10 participants who were not part of the investigated population on 2 separate occasions 24 hours apart. Participants were instructed not to participate in sport, dance activity or warm up during this 24 hour period to reduce the potential for ROM adaptations. Intra-rater reliability for total BS had an ICC of 0.96 indicating excellent reliability.

Localised JH, GJH spectrum disorder, peripheral JH

Localised JH was defined as a BS of 1 to 4¹ and GJH as a BS \geq 5¹ plus one or more secondary musculoskeletal manifestations of: (1) Musculoskeletal pain in two or more joints recurring daily for at least 3 months. (2) Chronic widespread pain for \geq 3 months. (3) Recurrent joint dislocation or joint instability in the absence of trauma (three or more atraumatic dislocations in the same joint or two or more atraumatic dislocations in two different joints occurring at different times or medical confirmation of joint instability at \geq 2 sites not related to trauma).¹ Peripheral JH is only present in the hands of feet¹ and in this study was assessed in the hands only as per the BS.²

hEDS

Clinical diagnosis of hEDS required the simultaneous presence of criteria 1, 2 and 3 which are composed of the following: (Criteria 1) GJH (BS \geq 5). Two or more from the following subsections (Criteria 2): (a) Systematic manifestations of generalised connective tissue disorder for which a total of five must be present which are outlined in table 2.¹⁰ (b) Positive family history with one or more first degree relatives meeting the criteria for hEDS. At least one of: (c) (i) Musculoskeletal pain in two or more joints recurring daily for at least 3 months. (c) (ii) Chronic widespread pain for \geq 3 months. (c) (iii) Recurrent joint dislocation or joint instability in the absence of trauma (three or more atraumatic dislocations in the same joint or two or more atraumatic dislocations in two different joints occurring at different times or medical confirmation of joint instability at \geq 2 sites not related to trauma).¹ (Criteria 3): Other connective tissue disorders such as lupus and rheumatoid arthritis must also be excluded.

For those features that for clinical diagnosis were outside the researchers clinical expertise (recurrent or multiple abdominal hernia, pelvic floor, rectal or uterine prolapse, dental crowding, mitral valve prolapse and aortic root dilation) participants were asked if they had suffered from these conditions and a positive family history for all criteria was determined via questioning. Skin hyperextensibility was measured via the pinching of the cutis on the volar surface of participants non-dominant forearm midway between the lateral epicondyle of the humerus and the distal head of the radius with the forearm supinated.³² The measurement was performed with skin callipers (Harpenden Skinfold Calliper, British Indicators, West Sussex, UK) by the researcher and a positive diagnosis was deemed as > 2cm.³² Abnormal scarring was defined as contusion based broad scars with thin wrinkled surface located on the extensor aspect of the elbows, knee and lower legs and was defined as present or absent.³³

Table 2 Clinical features of hEDS¹⁰

Clinical features
Skin extensibility
Unexplained striae
Bilateral piezogenic papules of the heel
Recurrent or multiple abdominal hernia
Atrophic scarring at ≥ 2 or more sites
Pelvic floor, rectal or uterine prolapse
Dental crowding or high or narrow palate
Positive bilateral wrist sign (Steinberg)
Positive bilateral thumb sign (Walker)
Arm span to height ≥ 1.05
Mitral valve prolapse
Aortic root dilation

Participants were provided with an adapted version of a validated and previously published questionnaire which had been used to score Brighton criteria responses.²⁴ This questionnaire determined the prevalence of hEDS criteria. To quantify questionnaire responses a positive response to a question was awarded 1 point and a negative response 0 points allowing a

maximum score of 17 points. The researcher was present to answer any queries during completion of the questionnaire.

Statistical analysis

The data satisfied the criteria for normal distribution and the total BS and total hEDS score were analysed using a one-way Anova to determine significance and a partial eta squared test (η^2) was used to calculate effect size. The assumptions of homogeneity of variance were determined using a Levene's test and were not met therefore a post hoc Games-Howell test was performed to identify significance differences between groups. All results were reported as means and standard deviations and significance was accepted as $P < 0.05$. Statistical analysis was performed using SPSS version 23 software (IBM Inc.)

Results

Localised JH, GJH prevalence and mean BS and hEDS scores

Table 3 summarises localised JH, GJH prevalence (total number and percentage of group value) and mean BS and hEDS scores. The number of participants who did not have any JH ranged from 50% (male rugby and male controls) to 1.59% (dancers). Localised JH (BS 1-4) ranged from 61.11% (netballers) to 28.57% (dancers). GJH (BS \geq 5) prevalence ranged from 69.84% (dancers) to 1.85% (male controls). Dancers demonstrated the highest BS (4.89 \pm 1.76) and hEDS criteria prevalence score (1.51 \pm 1.06) and male controls the lowest BS (1.07 \pm 2.17) and hEDS criteria prevalence score (0.53 \pm 0.68). There was a statistically significant difference between groups for BS ($F=29.001$, $p < 0.001$, $\eta^2=.28$) and post-hoc analysis revealed a significant difference between dancers and all other groups. Statistically significant findings are reported in table 4.

Table 3 Localised JH, GJH prevalence and mean BS and hEDS scores

CS	FR	MR	NP	FD	MC	FC
BS (0)	18 (24%)	27 (50%)	10 (13.89%)	1 (1.59%)	27 (50%)	24 (40%)
BS (1-4) Localised JH	43 (57.33%)	25 (46.30%)	44 (61.11%)	18 (28.57%)	26 (48.15%)	23 (38.33%)
BS (≥5) GJH	14 (18.67%)	2 (3.70%)	18 (25%)	44 (69.84%)	1 (1.85%)	13 (21.67%)
Mean BS	2.27 (2.24)	1.33 (1.97)	2.88 (2.03)	4.89 (1.76)	1.07 (1.55)	2.18 (2.17)
Mean hEDS	0.77 (0.89)	0.70 (0.88)	0.79 (0.82)	1.51 (1.06)	0.53 (0.68)	0.60 (0.79)

Abbreviations: FR; Female Rugby Players, MR; Male Rugby Players, NP; Netball Players, FD; Female Dancers, MC; Male Controls, FC; Female Controls; CS; Classification Score, BS Beighton Score, hEDS: hypermobility Ehlers-Danlos type

Table 4 Games-Howell post-hoc analysis of BS

Group		MD	95% CI	p value
FD	FR	2.62	1.64-3.60	< 0.001 [†]
	MR	3.56	2.55-4.57	< 0.001 [†]
	NP	2.01	1.06-2.96	< 0.001 [†]
	MC	3.82	2.94-4.69	< 0.001 [†]
	FC	2.71	1.67-3.74	< 0.001 [†]
FR	MC	1.20	0.24-2.15	0.005*
MR	NP	-1.55	-2.58- -0.50	0.001*
NP	MC	1.74	0.64- 2.83	0.001*
FC	MC	1.11	0.11-2.12	0.021*

[†] = $P < 0.001$

* = $P < 0.05$

Abbreviations: FR; Female Rugby Players, MR; Male Rugby Players, NP; Netball Players, FD; Female Dancers, MC; Male Controls, FC; Female Controls, MD; Mean Difference; CI; Confidence Intervals

hEDS prevalence

Five participants met the criteria for a clinical diagnosis of hEDS (1 female rugby player, 1 netballer and 3 dancers). There was a statistically significant difference for hEDS criteria prevalence as determined by one-way Anova ($F=10.31$, 375, $p < 0.001$, $\eta^2=.12$) and post-hoc analysis revealed a significant difference between dancers and all other groups. Statistically significant findings are reported in table 5.

Table 5 Games-Howell post-hoc analysis of hEDS criteria prevalence

Group		MD	95% CI	p value
FD	FR	0.73	0.25-1.22	< 0.001 [†]
	MR	0.80	0.28-1.32	< 0.001 [†]
	NP	0.72	0.24-1.19	< 0.001 [†]
	MC	0.98	0.51-1.45	< 0.001 [†]
	FC	0.91	0.42-1.39	< 0.001 [†]

[†] = $P < 0.001$

Abbreviations: FR; Female Rugby Players, MR; Male Rugby Players, NP; Netball Players, FD; Female Dancers, MC; Male Controls, FC; Female Controls, MD; Mean Difference; CI; Confidence Intervals

Table 6 summarises positive scores for hEDS criteria (total number and percentage of positive contribution). Within the dislocation/subluxation category, 46 positive scores were for dislocation and 4 for subluxation (2 male rugby, 2 female rugby). For abnormal skin all 6 positive scores were for skin hyperextensibility. With regard to the varicose veins/hernia/uterine and rectal prolapse category, 9 positive scores were for varicose veins and 3 for hernia (1 female rugby, 1 dancer, 1 rugby male).

Table 6 Positive score contribution to hEDS criteria

Group	Pain up to 3 m ≥ 2 JT	Chronic widespread pain ≥ 3 m	D SUB	MH	AS	VV HE UP RP	FH hEDS
FR	26 (34.66%)	3 (4%)	13 (17.3%)	0 (0%)	0 (0%)	3 (4.0%)	1 (1.33%)
MR	25 (46.30%)	3 (5.55%)	4 (7.41%)	1 (1.85%)	0 (0%)	4 (7.41%)	0 (0%)
NP	28 (38.88%)	1 (1.39%)	6 (8.33%)	1 (1.39%)	1 (1.39%)	1 (1.39%)	2 (2.78%)
FD	21 (33.33%)	7 (11.11%)	12 (19.05%)	0 (0%)	4 (6.35%)	2 (3.17%)	4 (6.35%)
MC	17 (31.48)	0 (0%)	6 (11.11%)	0 (0%)	1 (1.85%)	1 (1.85%)	0 (0%)
FC	10 (16.67%)	4 (6.67%)	9 (15%)	0 (0%)	0 (0%)	1 (1.66%)	1 (1.66%)

Abbreviations: FR; Female Rugby Players, MR; Male Rugby Players, NP; Netball Players, FD; Female Dancers, MC; Male Controls, FC; Female Controls, BS; Beighton Score, M; Month, JT; Joint(s), D; Dislocation, SUB; Subluxation, STR; Soft Tissue Rheumatism, MH; Marfanoid Habitus, Abnormal skin, ES; Eye signs, VV; Varicose Veins, HE; Hernia, UP; Uterine Prolapse, RP; Rectal Prolapse, hEDS; hypermobility Ehlers-Danlos type, FH; Family history

GJH and secondary musculoskeletal manifestations

Table 7 summarises the composition of secondary musculoskeletal manifestations in individuals diagnosed with GJH (total number and percentage of positive secondary musculoskeletal manifestation contribution). With regard to the dislocation/subluxation

subcategory, no participants suffered a subluxation. Dancers had the highest score for all categories.

Table 7 Composition of GJH and associated secondary musculoskeletal manifestations

Group	BS ≥5	Pain up to 3 m ≥ 2 JT	Chronic widespread pain ≥ 3 m	D SUB
FR	10 (100%)	7 (70%)	1 (10%)	6 (60%)
MR	2 (100%)	1 (50%)	2 (100%)	1 (50%)
NP	8 (80%)	7 (87.5%)	1 (12.5%)	2 (25%)
FD	19 (100%)	17 (89.47%)	2 (10.53%)	9 (47.37%)
MC	1 (100%)	1 (100%)	0 (0%)	0 (0%)
FC	7 (100%)	7 (100%)	0 (0%)	1 (14.29%)

Abbreviations: FR; Female Rugby Players, MR; Male Rugby Players, NP; Netball Players, FD; Female Dancers, MC; Male Controls, FC; Female Controls, BS; Beighton Score, M; Month, JT; Joint(s), D; Dislocation, SUB; Subluxation

Peripheral JH

Table 8 summarises the prevalence of peripheral JH (total number and percentage of group value in parenthesis). Rugby males had the highest JH prevalence (29.63%) with dancers the lowest (1.59%).

Table 8 Prevalence of peripheral JH

Group	LF	RF	LT	RT
FR 12 (16%)	11	7	4	3
MR 16 (29.63%)	8	8	11	8
NP 19 (26.39%)	17	14	5	5
FD 1 (1.59%)	1	1	1	1
MC 9 (15.79%)	9	6	4	0
FC 7 (11.67%)	6	4	7	5

Abbreviations: FR; Female Rugby Players, MR; Male Rugby Players, NP; Netball Players, FD; Female Dancers, MC; Male Controls, FC; Female Controls; LF; Left 5th metacarpophalangeal joint, RF; Right 5th metacarpophalangeal joint, LT; Left thumb, RT; Right thumb

Discussion

With regard to the primary aim of this study localised JH (BS 1-4) ranged from 61.11% (netballers), 57.33% (female rugby), 48.15% (male controls), 46.30% (male rugby), 38.33% (female controls) to 28.57% (dancers) (table 3). Male rugby and male controls demonstrated the highest prevalence of no hypermobile joints (50%) with dance the lowest (1.59%). Comparison of localised JH with previous literature is difficult as no studies have reported BS in this manner of 1-4 grading since the introduction of the new hypermobility framework.¹ However, it is apparent that there is a greater prevalence of no hypermobile joints in males and comparison between female rugby and male rugby demonstrated that no JH prevalence was

doubled in males and also greater in male than female controls suggesting a gender consideration. Dancers demonstrated the lowest localised JH due to the greater prevalence of $BS \geq 5$.

GJH ($BS \geq 5$) prevalence ranged from 69.94% (dancers), 25% (netballers), 21.67% (female controls), 18.67% (female rugby), 3.70% (male rugby) to 1.85% (male controls) (table 3). In dancers, GJH values were higher than the 66%⁸ and 57% in ballet students³⁴ and lower in netballers than the 66%²⁰ previously reported. Male rugby values were lower than previously reported 24%¹⁵ and 20%³⁵. The values for male controls and female controls were lower than previous findings in male students of 36.7% but higher than the 13.7% in female students in a study utilising $GJH \geq 5$.³⁶ Comparison with studies^{8,15,20,34,35} is limited by their use of the previously recommended cut off point of $BS \geq 4$.

Total BS was significantly greater in dancers in comparison to all other groups and significant differences existed between female rugby and male controls, netballers and male rugby, netballers and male controls and female controls and male controls (table 4) and demonstrated a large effect size. The significant findings within dancers may highlight a potential performance adaptation and/or selection of individuals with JH. Significant findings between control groups suggests a potential gender effect which supports previous findings of a higher prevalence of JH in females.¹³ Between rugby females, netballers, dancers and male subgroups a gender difference may also have existed. The finding of no significance between the two male subgroups may suggest that rugby males do not demonstrate a JH adaptation.

The mean male rugby BS of 1.33 was lower than previous findings of 2.0¹⁵ and 1.62³⁵ and the mean netball BS of 2.88 (table 3) was lower than 3.99¹⁶ and 3.96²⁰ which may reflect the different sporting levels and participant ages. The mean BS for female rugby of 2.27 was similar to the 2.60 previously reported²⁴ and as these values were similar to female controls it suggests a JH performance adaptation does not occur. Mean dancers BS was 4.89 and was greater than the mean BS of 4.36 previously reported in dancers.^{18,26} Determination of the BS is important within dance as within ballet an increased injury risk comes from the level of muscular effort required by hypermobile dancers to maintain stability.³⁷ Hypermobile individuals stand with hyperextended joints making maintenance of a position difficult³⁸ and fatigue levels are higher in hypermobile dancers⁸ and neuromuscular control, proprioception and functional stability are impaired after fatigue.³⁹ Therefore there may be a need for

appropriate training programmes which consider work:rest ratios and potentially may reduce injury risk.

With regard to the secondary aim of the study and the prevalence of secondary musculoskeletal manifestations required for a positive diagnosis of GJH, pain up to 3 months in 2 or more joints and dislocation/subluxation was most prevalent in dancers (table 7). This could potentially be attributed to the extreme ranges of motion required in dance which may potentially increase the risk of dislocation/subluxation. Female rugby had the second highest numbers of dislocations/subluxations. $BS \geq 5$ was the greatest contributor in all groups and chronic widespread pain for more than 3 months was most prevalent in dancers and male rugby however the frequency of this factor across all groups ranged from 0 to 2 highlighting that it was not a dominant factor. Five females met the criteria for clinical diagnosis of hEDS which included 3 dancers. All five had a $BS \geq 5$, secondary musculoskeletal manifestations and a positive family history. No participants in the cohort met 5 or more criteria for systematic manifestations of generalised connective tissue disorder. No males met the criteria due to the lower BS scores, and lack of systematic manifestations of generalised connective tissue disorder and a positive family history.

The study awarded points for the presence of criteria associated with hEDS utilising an adapted scoring system which assessed the presence of Brighton criteria signs and symptoms.²⁴ There was a significant difference between the prevalence of hEDS criteria in dancers and all other groups and a large effect size was demonstrated. Mean hEDS scores were greatest in dancers (1.51) with male controls demonstrating the lowest score (0.53). There was a limited range in total hEDS criteria score with the highest score 5/17 achieved in dancers. With regard to the number of positive findings for hEDS type (table 6), pain up to 3 months in ≥ 2 joints was most prevalent in male rugby (46.30%), netballers (38.88%), female rugby (34.66%) and dancers (33.33%). This high prevalence in male rugby may reflect the increased risk of injury associated with contact sports. Dislocation was most prevalent in dancers (19.05%), and female rugby (17.3%). Previous research has reported that Glenohumeral joint instability was nearly 2.5 times more likely in participants with a $BS \geq 2$ ⁴⁰ and generalised ligamentous laxity was more common in individuals with a primary traumatic anterior shoulder dislocation.⁴¹ Male controls had the lowest number of dislocations/subluxations. Chronic widespread pain (19.05%) and abnormal skin (6.35%) was highest in dancers and this may reflect that many dancers continue to dance while in pain and therefore not providing injuries with an opportunity

to heal. Varicose veins/hernia/uterine or rectal prolapse were most prevalent in male rugby (7.41%), female rugby (4%) and dancers (3.17%). The absence of marfanoid habitus in dancers is unusual but may reflect the amateur level of the dancers and within a professional dance company this physical presentation may be considered an attribute. The prevalence of marfanoid habitus in netballers (1.39%) may be considered low as this tall, long limbed physique may be considered beneficial for shooters and goalkeepers. As the hypermobility spectrum¹ is relatively new direct comparison of the prevalence of certain criteria that compose secondary musculoskeletal manifestations and hEDS is difficult with most studies utilising the Brighton criteria. In netballers a prevalence of abnormal skin (19%) and arthralgia > 3 months (15%) were reported²⁰ and in ballet students, excluding the BS criteria the most prevalent Brighton criteria was arthralgia (27.3%) and dislocation (22.7%)³⁴ which replicates the order in dancers within this study.

The prevalence of GJH among dancers is related to age, gender and ethnicity^{42,43} and there is a need to consider how these factors may interact and contribute to injury risk and performance factors. Previous research has demonstrated that hypermobility associated with collagen structure is related to skin elasticity and hernias, while genetic factors contribute to JH in individuals with congenital dislocation and spinal scoliosis.⁴⁴ The GJH assessment outlined in the methodology could be used as an initial screen before a more detailed medical examination is performed to clarify the potential relationship all variations of the hypermobility spectrum and these medical conditions. Previous research highlighted increased injury prevalence with JHS as defined by the Brighton criteria but not within JH as defined by the BS.⁴⁵ Although the Brighton criteria is no longer advocated it is important that clinicians consider the full hypermobility spectrum to enhance understanding of the individuals they are working with and not to solely rely on the BS.

Peripheral JH was most prevalent in male rugby (29.63%), netball (26.39%) and least prevalent in female dancers (1.59%) (table 8). Comparison with previous literature is not possible due to peripheral JH been part of the new hypermobility spectrum however the high prevalence in male rugby is likely due to the generally low BS while in contrast the low prevalence in dancers is due to the higher BS at a greater variety of joint locations. Peripheral JH within netballers may reflect a performance adaptation as an increased range of motion at the fingers and thumbs may allow improved performance when reaching for the ball however this requires further investigation.

The high prevalence of JH in dancers provides two possibilities, firstly that dance results in individuals developing JH and secondly that dance favours the selection of JH individuals. Long term prospective studies throughout a dancers career from childhood are required to investigate this. JH classification in rugby union, netball and dance suggests that the consideration of gender, sport and dance participation is important in determining normal values and there is a need to consider age, gender and ethnicity.⁴⁴ The groups in this study have varying levels of physical demands and injury risk including the contact demands of rugby and non-contact demands of netball and dance. Clinical decisions regarding injury prevention, training load and sport selection based on JH should consider the gender and predominant activity of the individual so that the BS can be interpreted and related to expected values within their domain. This information could be combined with a full musculoskeletal screening to aid development of training and injury prevention programmes. Within the study some limitations must be acknowledged namely that the findings are limited to the populations investigated and the BS does not report specific joint ROM. Further studies should consider male dancers and report joint ROM to allow for greater investigation of specific ROM values.

Conclusion

This study is the first to examine the prevalence of localised JH, GJH, hEDS and peripheral JH in rugby, netball and dance in relation to the hypermobility spectrum¹ and has highlighted that the BS is significantly greater in female dancers than in rugby, netball, male and female controls which may be suggestive of an activity adaptation or a selection of JH individuals. Gender differences existed for the BS with values significantly greater in female controls than male controls and between all other female subgroups and male subgroups. GJH and the criteria associated with hEDS is more prevalent in dancers than rugby, netball and male and female controls. The higher prevalence of dislocation/subluxation in dancers may represent the ROM demands of aesthetic performance. Localised JH and peripheral JH is least prevalent in dancers due to the higher number of hypermobile joints in comparison to other groups. The prevalence of hEDS was low with only 5 females diagnosed with this condition. The initial implementation of the BS and hypermobility spectrum was as an epidemiological tool and not as a sport or dance specific tool and therefore the development of sport or dance specific grading scales based on normal values for each group seems a logical progression which could be used to manage performance demands and injury risk.

References

1. Castori M, Tinkle B, Levy H, Grahame R, Malfait F, Hakim A. A framework for the classification of joint hypermobility and related conditions *Am J Med Genet.*2017;175C: 148-157.
2. Beighton P, Solomon L, Soskolne CL. Articular mobility in an African population. *Ann Rheum Dis.*1973;32:413-18.
3. Remvig L, Jensen DV, Ward RC. Are diagnostic criteria for general joint hypermobility and benign joint hypermobility based on reproducible and valid tests? A review of the literature. *J Rheumatol.*2007;34:798-803.
4. Remvig L, Flycht L, Christensen KB, Juul-Kristensen B. Lack of consensus on tests and criteria for generalised joint hypermobility, Ehlers-Danlos syndrome: hypermobile type and joint hypermobility syndrome. *Am J Med Genet A.*2014; 164a:591-596.
5. Simpson MR. Benign Joint Hypermobility Syndrome: Evaluation, Diagnosis, and Management. *J Am Osteopath Assoc.*2006;106:531-536.
6. Grahame R, Bird HA, Child A. The revised (Brighton 1998) criteria for the diagnosis of benign joint hypermobility syndrome (BJHS). *J Rheumatol.*2000;27:1777-1779.
7. Hakim A, Grahame R. Joint hypermobility. *Best Pract Res Clin Rheumatol.*2003;17:989-1004.
8. Scheper MC, de Vries JE, de Vos R, Verbunt J, Nollet F, Engelbert RHH. Generalised joint hypermobility in professional dancers: a sign of talent or vulnerability? *Rheumatology* 2013;52:651-658.
9. Tinkle BT, Bird HA, Grahame R, Lavellee M, Levy HP, Silience D. The lack of clinical distinction between the hypermobility type of Ehlers-Danlos syndrome and the joint hypermobility syndrome (a.k.a. hypermobility syndrome). *Am J Med Genet.*2009;Part A 149a:2368-2370.
10. Malfait F, Francomano C, Byers P, Belmont J, Berglund B, Black J et al. The 2017 International Classification of the Ehlers-Danlos syndromes. *Am J Med Genet Part C Semin Med Genet.*2017;175C:8-26.
11. Beighton P, de Paepe A, Danks D, Finidori G, Gedde-Dahl T, Goodman R, Hall JG, Hollister DW, Horton W, McKusick VA et al. International nosology of heritable disorders of connective tissue, Berlin, 1986. *Am J Med Genet.*1988;29:581-594.
12. Seckin U, Tur BS, Yilmaz O, Yagci I, Bodur H, Arasil T. The prevalence of joint hypermobility among high school students. *Rheumatol Int.*2005;25: 260-263.

13. Simmonds JV, Keer RJ. Hypermobility and the hypermobility syndrome. *Man Ther.* 2007;12:298-309.
14. Klemp P, Williams SM, Stansfield SA. Articular mobility in Maori and European New Zealanders. *Rheumatology (Oxford)*.2002;41 (5):554-557.
15. Stewart S, Burden S. Does generalised ligamentous laxity increase seasonal incidence of injuries in male first division club rugby players? *Br J Sports Med*.2004;38:457-460.
16. Smith R, Damodaran A, Swaminathan S, Campbell R, Barnsley L. Hypermobility and sports injuries in junior netball players. *Br J Sports Med*.2005;39:628-631.
17. Klemp P, Stevens JE, Isaacs SA. Hypermobility study in ballet dancers. *J Rheumatol*. 1984;11 (5):692-696.
18. Bronner S, Bauer NG. Risk factors for musculoskeletal injury in elite pre-professional modern dancers: A prospective cohort prognostic study. *Phys Ther Sport*.2018;31:42-51.
19. Pacey V, Nicholson LL, Adams RD, Munn, J, Munns CF. Generalised joint hypermobility and risk of lower limb joint injury during sport. A systematic review with meta-analysis. *Am Journal Sports Med*.2010;38:1487-1497.
20. Soper K, Simmonds J, Kaz H, Ninis N. The influence of joint hypermobility on functional movement control in an elite netball population: A preliminary cohort study. *Phys Ther Sport*.2015;16:127-134.
21. McCormack M, Briggs J, Hakim A, Grahame R. Joint laxity and the benign joint hypermobility syndrome in student and professional ballet dancers. *J Rheumatol*.2004;31: 173-178.
22. Briggs J, McCormack M, Hakim AJ, Grahame R. Injury and Joint Hypermobility syndrome in ballet dancers-a 5 year follow-up. *Rheumatology*.2009;48:1613-1614.
23. Rombaut L, Malfait F, De Wandele I, Thijs Y, Palmas T, De Paepe A, Calders P. Balance, gait, falls, and fear of falling in women with hypermobility type of Ehlers-Danlos syndrome. *Arthritis Care Res*.2011;63:1432-1439.
24. Armstrong R, Greig M. The Beighton score as a predictor of Brighton criteria in Sports and Dance. *Phys Ther Sport*. 2018.32;145-154.
25. Finsterbush A, Pogrund H. The hypermobility syndrome: Musculoskeletal complaints in 100 consecutive cases of generalised joint hypermobility. *Clin Orthop*.1982;168:124-27.
26. Gannon LM, Bird HA. The quantification of joint laxity in dancers and gymnasts. *J Sport Sci*.1999;17:743-750.
27. Graham R, Jenkins JM. Joint hypermobility: an asset or a liability? A study of joint mobility in ballet dancers. *Ann Rheum Dis*.1972;31:109-111.

28. Chorba RS, Chorba DJ, Bouillon LE, Overmyer CA, Landis JA. Use of a functional movement screening tool to determine injury risk in female collegiate athletes. *N Am J Sports Physical Ther.* 2010;5:47–54.
29. Bird HA. Rheumatological aspects of dance. *J Rheumatol.* 2004;31:12-13.
30. Juul-Kristensen B, Røgind H, Jensen DV, Remvig L. Inter-examiner reproducibility of tests and criteria for generalized joint hypermobility and benign joint hypermobility syndrome. *Rheumatology.* 2007;46:1835-1841.
31. Weir JP. Quantifying test-retest reliability using the intraclass correlation coefficient and the SEM. *Journal Strength Cond Res.* 2005;19: 231-240.
32. Remvig L, Duhn PH, Ullman S, Kobayasi T, Hansen B, Juul-Kristensen B, Jurvelin JS, Arokosi. Skin hyperextensibility and consistency in patients with Ehlers-Danlos syndrome and benign joint hypermobility syndrome. *Scand J Rheumatol.* 2009;38 (3):227-230.
33. Remvig, L, Duhn, PH, Ullman, S, Arokoski, J, Jurvelin, J, Safi, A et al. Skin signs in Ehlers–Danlos syndrome: clinical tests and para-clinical methods, *Scand J Rheumatol.* 2010; 39 (6):511-517.
34. Sanches SB, Oliveira GM, Osório FL, Crippa JAS, Martín-Santos R. Hypermobility and joint hypermobility syndrome in Brazilian students and teachers of ballet dance. *Rheumatol Int.* 2015;35:741-747.
35. Oddy C, Johnson MI, Jones G. The effect of generalised joint hypermobility on rate, risk and frequency of injury in male university-level rugby league players: a prospective cohort study. *BMJ Open Sport Exerc Med.* 2016;1:e000177. doi:10.1136.
36. Russek LN, Errico DM. Prevalence, injury rate and, symptom frequency in generalized joint laxity and joint hypermobility syndrome in a “healthy” college population. *Clin Rheumatol.* 2016;35:1029-1039.
37. Foley EC, Bird HA. Hypermobility in dance: asset, not liability. *Clin Rheumatol.* 2013; 32:455-461.
38. Keer R, Butler K. Physiotherapy and occupational therapy in the hypermobile adult. In: Hakim AJ, Keer R, Graham R (eds) *Hypermobility, fibromyalgia and chronic pain.* 1st Edition. Churchill Livingstone, Elsevier, London. pp143-161. 2010.
39. Ribeiro F, Santos F, Gonclaves P, Oliveira J. Effects of volleyball match-induced fatigue on knee joint position sense. *Eur J Sport Sci.* 2008;8:397-402.
40. Cameron KL, Duffey ML, DeBerardino TM, Stoneman PD, Jones CJ, Owens BD. Association of generalized joint hypermobility with a history of glenohumeral joint instability. *Journal of Athletic Training.* 2010;45:253–258.

41. Chahal J, Leiter J, McKee M D, Whelan DB. Generalized ligamentous laxity as a predisposing factor for primary traumatic anterior shoulder dislocation. *J Shoulder Elbow Surg.* 2010;19:1238–1242.
42. Larsson LG, Baum J, Mudholkar GS. Hypermobility: features and differential incidence between the sexes. *Arthritis Rheum.* 1987;30:1426-1430.
43. Rikken-Bultman DG, Wellink L, van Dongen P. Hypermobility in two Dutch School populations. *Eur J Obstet Gynecol Reprod Biol.* 1997;73:189-192.
44. Juul-Kristensen, B, Kristensen, JH, Frausing, B, Jensen DV, Røgind H, Remvig L. Motor competence and physical activity in 8-year-old school children with generalized joint hypermobility. *Pediatrics.* 2009;124:1380-1387.
45. Ruemper A and Watkins K. Correlations between general joint hypermobility and joint hypermobility syndrome and injury in contemporary dance students. *J Dance Med Sci.* 2012; 16:161-166.