

# Generalized joint hypermobility in professional dancers: a sign of talent or vulnerability?

Mark C. Scheper<sup>1,2</sup>, Janneke E. de Vries<sup>1,2</sup>, Rien de Vos<sup>3</sup>, Jeanine Verbunt<sup>4</sup>, Frans Nollet<sup>2</sup> and Raoul H. H. Engelbert<sup>1,2</sup>

## Abstract

**Objective.** To study the impact of generalized joint hypermobility (GJH) in professional dancers on physical fitness, musculoskeletal complaints and psychological distress.

**Methods.** Thirty-six professional dancers were recruited and compared with control subjects (mean age 20.1, range 17–27). Height, weight, Beighton score, physical fitness (walking distance, muscle strength, estimated VO<sub>2</sub>max), musculoskeletal complaints (pain, fatigue) and psychological distress (anxiety, depression) were measured.

**Results.** Univariate analysis revealed, in between-group analysis, that dancers (with and without GJH) had higher physical fitness [the six-minute walk test (6MWT):  $\Delta D = +8.4\%$ ,  $P = 0.001$ ; VO<sub>2</sub>max:  $\Delta D = +12.8\%$ ,  $P = 0.01$ ], fatigue (checklist individual strength:  $\Delta D = +80.3\%$ ,  $P < 0.0001$ ) and greater psychological distress (Hospital Anxiety and Depression Scale:  $\Delta D = +115.0\%$ ,  $P < 0.0001$ ). When comparing dancers and control subjects with GJH to those without GJH, lower levels of physical fitness (muscle strength:  $\Delta D = -11.3\%$ ,  $P < 0.0001$ ; 6MWT:  $\Delta D = -9.9\%$ ,  $P < 0.0001$ ), more fatigue (checklist individual strength:  $\Delta D = +84.4\%$ ,  $P < 0.0001$ ) and greater psychological distress (Hospital Anxiety and Depression Scale:  $\Delta D = +79.6\%$ ,  $P < 0.0001$ ) were observed in subjects with GJH. Multivariate analysis showed that dancers have higher levels of physical fitness (6MWT,  $P = 0.001$ ; VO<sub>2</sub>max,  $P = 0.020$ ); however, when taking GJH into account, this advantage disappeared, indicating lower levels of physical fitness in comparison with control subjects (6MWT,  $P = 0.001$ ; muscle strength,  $P < 0.0001$ ; VO<sub>2</sub>max,  $P = 0.040$ ). Dancers experienced more fatigue ( $P = 0.001$ ) and psychological distress ( $P < 0.0001$ ). This was associated with even more fatigue ( $P = 0.010$ ) and psychological distress ( $P = 0.040$ ) when GJH was present.

**Conclusion.** Dancers with GJH seem more vulnerable to musculoskeletal and psychological complaints. In addition, GJH was also associated with lower physical fitness, despite training. Caregivers for professional dancers should monitor closely the physical capabilities and the amount of psychological strain.

**Key word:** generalized joint hypermobility, dance, physical fitness, musculoskeletal complaints, psychological distress.

## Introduction

Within the (professional) dance community, the presence of generalized joint hypermobility (GJH) is regarded as an important feature in reaching the level of a performing dancer [1], and it is often used as a measure for selecting the most promising students. For aesthetic reasons, increased flexibility is often promoted and is viewed as an essential integral part of professional dance education. The prevalence of GJH among dancers varies between 11% and 97% [2] and is age, gender and ethnicity dependent [3, 4]. This percentage of dancers

<sup>1</sup>Education of Physiotherapy, Amsterdam School of Health Professions, University of Applied Sciences, Amsterdam, <sup>2</sup>Department of Rehabilitation, Academic Medical Center, University of Amsterdam, Amsterdam, <sup>3</sup>Department of Clinical Epidemiology and Biostatistics, Academic Medical Center, University of Amsterdam, Amsterdam and <sup>4</sup>Department of Rehabilitation Medicine, School for Public Health and Primary Care: CAPHRI, Maastricht University Medical Center, Maastricht, The Netherlands.

Submitted 13 April 2012; revised version accepted 6 July 2012.

Correspondence to: Mark C. Scheper, Amsterdam School of Health Professions, Tafelbergweg 51, 1105BD, Amsterdam, The Netherlands. E-mail: m.c.scheper@hva.nl

classified with GJH exceeds the prevalence of GJH in the general population, ranging from 0.6% to 31.5% [3–5]. Although for dancers flexible joints are considered to be aesthetically beneficial, for others GJH can be potentially disabling [1].

GJH is also a common feature of hereditary diseases of connective tissue, like osteogenesis imperfecta, and the Ehlers–Danlos and Marfan syndromes [6]. In these disorders, GJH is often accompanied by morphological features, joint dislocations, bone fragility and hyperextensible skin. Genetic alterations have been documented that account for the changed structural components of connective tissue [7]. However, previous research has also identified an additional class of disorders sharing the presence of increased connective tissue laxity, but without clear biological markers—hypermobility syndrome and Ehlers–Danlos syndrome, hypermobile type [8]. Individuals diagnosed with the above-mentioned disorders frequently experience severe musculoskeletal complaints in terms of pain and fatigue that often lead to loss of functional ability [9, 10] and impairments in daily life [11].

The literature reports that individuals with symptomatic forms of GJH often have less physical fitness (strength, stamina) [12], loss of proprioceptive acuity [13], autonomic dysfunction [14] and increased levels of depression and anxiety [15]. The literature regarding dance-specific injuries is scarce and heterogeneous because of the considerable variation in dance styles (e.g. classical ballet vs break-dancing) and differences in professionalism that have their own unique physical requirements [2]. In dancers, high incidence rates of musculoskeletal injury have been reported, for the majority in the lower extremities and back, predominated by soft tissue lesions and overuse injuries [16]. Various potential risk factors for dancers have been suggested, ranging from physical overload to psychological distress; however, conclusive evidence for any of the reported risk factors is lacking [17]. Recently a study by McCormack *et al.* [18] demonstrated that the incidence of symptomatic GJH (hypermobility syndrome) is about four times more frequent in dancers when compared with age- and gender-matched control subjects. Despite these data, it remains unknown what the impact of GJH in professional dancers is and whether GJH affects dancers differently when compared with the normal population. Therefore the objective of the present study is to determine the impact of GJH in dancers on functional ability, physical fitness, musculoskeletal complaints and psychological functioning in comparison with matched control subjects.

## Methods

### Participants

In this cross-sectional study, with convenience sampling, participants were recruited from the Amsterdam School of Arts Academy for dance and theatre in their final year of dance education. All dancers in their final year of formal dance education were invited to participate. Dancers were compared with age- and gender-matched volunteers

recruited from the Amsterdam School of Health Professions, The Netherlands, to serve as control subjects. Because of the small sample of male dancers willing to participate in the current study ( $n=2$ ), the study was limited to female dancers only. The study was carried out between May 2011 and July 2011 at the outpatient clinic of the Department of Rehabilitation at the Academic Medical Center Amsterdam, The Netherlands. Subjects were eligible for inclusion when (i) no orthopaedic, cardiopulmonary, rheumatological or neurological conditions or disorders influencing physical performance were present, and (ii) they were able to understand the questionnaires or to adhere to the protocol.

### Descriptives

Demographic data were collected regarding age, gender, height and weight. Standing height and weight were measured without heavy clothing and shoes to the nearest centimetre and 100 g using a standardized method. BMI was calculated with the formula weight divided by height squared ( $\text{kg/m}^2$ ). Physical activity level (PAL) was determined according to the Short Questionnaire to Assess Health Enhancing Physical Activity (SQUASH) [19], and it was converted into metabolic equivalent tasks according to the compendium of physical activities [20].

The presence of joint (hyper)mobility was quantified by the Beighton score. For the Beighton score, the protocol by Juul-Kristensen *et al.* [21] was adapted to guarantee uniformity. In all included subjects, the Beighton score was determined before the measurements without a warming-up phase. A participant was classified with GJH when a Beighton score of  $\geq 4$  was obtained. Written informed consent was obtained from all participants according to the Declaration of Helsinki. The study was approved by the Medical Ethics Board of the Academic Medical Center, Amsterdam, The Netherlands.

### Outcomes

Outcomes were classified into the following three domains: physical fitness (functional walking ability, muscle strength, estimated  $\text{VO}_2\text{max}$ ), musculoskeletal complaints (pain intensity, fatigue) and psychological distress (anxiety, depression and combined scores).

#### Physical fitness

The six-minute walk test (6MWT) was applied as a walking capacity measure according to the International Classification of Functioning [22]. The test was performed on an 8-m track in a straight corridor as described by Gulmans *et al.* [23]. Participants were instructed to cover the largest possible distance in 6 min at a self-chosen walking speed. Turns were made on both ends of the 8-m track. The distance walked was recorded with a lap counter. Each time the patient returned to the starting line, the lap counter was clicked once. Patients were encouraged every minute in a standardized way, recorded with a stopwatch. At the end of the test, the patient was asked to stand still and the distance covered in the final partial

lap was measured. This was quantified with a measuring tape.

Muscle strength of proximal and distal muscles in the lower and upper extremities was measured bilaterally in a standardized way [12] with a hand-held dynamometer (Citec, Groningen, The Netherlands). Measurements were consecutively performed three times and the highest value was registered. In the upper extremity, shoulder abductors and grip strength were measured. In the lower extremity, hip flexors, knee extensors and dorsal extensors of the foot were measured. All measurements were performed according to the break method [12], with the exception of knee extension and grip strength. For these measurements, the make method was applied because of the inability of the assessors to break the generated force of the participant [24]. A total muscle strength index was calculated by summation of all individual muscles (left and right).

Submaximal exercise capacity was measured with the submaximal modified Harvard Step Test. It is a commonly used and valid method [25] to estimate  $\text{VO}_2\text{max}$ . Participants were instructed to step on and off a bench of standardized height (males 40 cm, females 33 cm) and at a standard rhythm (45 bpm) for 6 min. Continuous heart frequency monitoring (Hf) was applied (Polar, team 2 monitoring system). The heart rate of the 5th and 6th minutes were averaged and used to determine the estimated  $\text{VO}_2\text{max}$  based on the Åstrand Rhythmic nomogram [26].  $\text{VO}_2\text{max}$  was expressed in litres/kilogramme/minute.

#### Musculoskeletal complaints

Pain was quantified according to the visual analogue scale expressed in millimetres, ranging from no pain at all (score 0 mm) to worst pain ever experienced (score 100 mm). Subjects rated the pain intensity they perceived in the previous 2 weeks.

Fatigue was quantified by the checklist individual strength (CIS). The CIS measures four dimensions of fatigue: subjective experience of fatigue, and reduction in motivation, activity and concentration. The CIS was reported to be reliable and valid in healthy control subjects and patients diagnosed with chronic fatigue syndrome and other chronic diseases [27]. The total CIS score was calculated through summation of all subitems, resulting in a score ranging from 0 to 100, and this score was used for analysis [27].

#### Psychological distress

Psychological distress was measured with the Hospital Anxiety and Depression Scale (HADS). The HADS is a 14-item self-report screening scale that was originally developed to indicate the possible presence of anxiety and depressive states in the setting of a medical outpatient clinic [28]. It contains two 7-item scales: one for anxiety and one for depression, both with a score range of 0–21. The psychometric properties of the HADS have been well established and have been extensively used in clinical practice and research [28]. Total scores and item scores were used for analysis.

#### Statistics

Descriptive statistics were used to describe all relevant variables. Normality of data was checked using the Kolmogorov–Smirnov test. Normally distributed data were expressed as mean and standard deviation, skewed data were presented as median and percentiles (P50, P25–P75).

To establish differences between groups (dancer vs non-dancer) and to determine the effect of GJH (Beighton  $\geq 4$ ), an independent *t*-test was applied. To determine the contributions of each factor and to control for confounding, multiple linear regression models were constructed for each outcome. In these models, the primary outcomes (physical fitness, musculoskeletal complaints and psychological distress) were used as dependent variables. Group (dancer: Yes/No) and classification GJH (Yes/No) were dichotomized (1/0) and used as independent variables, corrected for potential confounders (BMI and PAL). Data are expressed as regression coefficient (B), with corresponding standard error (s.e) and 95% CI. Results were considered to be statistically significant at a *P*-level of  $<0.05$ . All analyses were performed in SPSS 16.0 (SPSS, Inc., Chicago, IL, USA).

#### Results

All initially invited subjects were willing to participate, and all fulfilled the inclusion criteria and were included in the analysis ( $n = 72$ : 36/36). The presence of GJH was significantly higher among dancers (66%) in comparison with controls (29%) ( $\chi^2 = 12.995$ ,  $P = 0.001$ ). The distribution of the Beighton scores over groups is provided in Table 1. An overview of the included study population is provided in Table 2.

#### Univariate analysis

Univariate analysis revealed that dancers in comparison with controls had significantly lower BMI (BMI:  $\Delta D = -7.4\%$ ,  $T = -2.991$ ,  $P = 0.004$ ), greater walking capacity (6MWT:  $\Delta D = +8.4\%$ ,  $T = 3.319$ ,  $P = 0.001$ ), higher estimated  $\text{VO}_2\text{max}$  ( $\text{VO}_2\text{max}$ :  $\Delta D = +12.8\%$ ,  $T = 2.586$ ,  $P = 0.012$ ), were more fatigued (CIS:  $\Delta D = +80.3\%$ ,  $T = 4.885$ ,  $P < 0.0001$ ) and experienced more psychological distress (HADS:  $\Delta D = +115.0\%$ ,  $T = 4.823$ ,  $P < 0.0001$ ) for anxiety (HADS-A:  $\Delta D = +105.4\%$ ,  $T = 4.606$ ,  $P < 0.0001$ ) and depression (HADS-D:  $\Delta D = +143.7\%$ ,  $T = 4.061$ ,  $P < 0.0001$ ). For the remaining outcomes, no significant differences between dancers and control subjects were found.

When comparing the total population of subjects classified with or without GJH, subjects with GJH had significantly lower functional walking distance (6MWT:  $\Delta D = -9.9\%$ ,  $T = -14.076$ ,  $P < 0.0001$ ) and lower muscle strength (muscle strength:  $\Delta D = -11.3\%$ ,  $T = -3.927$ ,  $P < 0.0001$ ), whereas more fatigue (CIS:  $\Delta D = +84.4\%$ ,  $T = 4.401$ ,  $P < 0.0001$ ) and higher levels of total psychological distress (HADS:  $\Delta D = +79.6\%$ ,  $T = 3.626$ ,  $P = 0.001$ ) as well as anxiety (HADS-A:  $\Delta D = +77.2\%$ ,  $T = 3.618$ ,  $P = 0.001$ ) and depression (HADS-D:  $\Delta D = +45.6\%$ ,

**TABLE 1** Clinical characteristics of the included population

Outcomes	Control subjects				Dancers			
	Non-GJH (Beighton <4) (n = 27)		GJH (Beighton ≥4) (n = 9)		Non-GJH (Beighton <4) (n = 12)		GJH (Beighton ≥4) (n = 24)	
	Mean (s.d.)	Range	Mean (s.d.)	Range	Mean (s.d.)	Range	Mean (s.d.)	Range
Subject characteristics								
Age, years	20 (2)	17–27	20 (2)	17–24	20 (3)	17–27	20 (2)	17–24
BMI <sup>a</sup>	22.2 (2.6)	18.8–28.4	22.2 (2.2)	18.7–25.5	20.6 (2.2)	16.8–23.5	20.8 (1.7)	17.9–23.1
Total PAL (METs)	169.5 (90.0)	24.2–391.2	176.5 (104.3)	85.4–435.2	206.2 (110.4)	71.1–491.5	192.3 (101.1)	46.2–411.5
Physical fitness								
6MWT, m <sup>a,b</sup>	604.6 (38.0)	543.0–708.0	477.7 (47.2)	418.5–572.0	662.8 (42.2)	614.2–780.0	599.9 (45.8)	492.0–706.0
Muscle strength, N <sup>b</sup>	2000.2 (214.8)	1751.3–2572.7	1803.8 (103.5)	1651.7–1960.3	2081.8 (252.8)	1740.3–2638.0	1826.4 (243.7)	1387.7–2373.7
VO <sub>2</sub> max, l/kg/min <sup>a</sup>	39.9 (7.0)	30.2–54.3	36.1 (7.1)	29.6–53.1	47.1 (11.3)	35.5–79.1	42.4 (7.6)	29.7–59.7
Musculoskeletal complaints								
Pain, mm	42.4 (21.3)	0.0–78.7	55.7 (12.8)	33.0–79.0	33.7 (19.6)	0.0–59.6	41.0 (18.5)	11.0–72.0
Fatigue, CIS <sup>a</sup>	22 (17)	5–72	42 (24)	15–97	43 (17)	10–75	51 (17)	15–83
Psychological function								
Depression, HADS <sup>a,b</sup>	2 (2)	0–9	3 (4)	0–12	4 (3)	0–10	5 (3)	1–11
Anxiety, HADS <sup>a,b</sup>	3 (3)	0–12	6 (3)	2–10	7 (4)	1–15	7 (4)	2–15
Total, HADS <sup>a,b</sup>	5 (5)	1–18	9 (6)	3–22	11 (7)	1–25	13 (6)	3–26

MET, metabolic equivalent task. <sup>a</sup>Indicates a statistically significant effect of group (dancer vs non-dancer). <sup>b</sup>Indicates statistically significant effects of GJH (yes/no)

**TABLE 2** Distribution Beighton score over joints for each group

Beighton items	Dancers		Control subjects	
	Percentage positive (n)	Percentage negative (n)	Percentage positive (n)	Percentage negative (n)
Thumb				
Left	61.1 (22)	39.9 (14)	41.7 (15)	58.3 (21)
Right	61.1 (22)	39.9 (14)	41.7 (15)	58.3 (21)
Little finger				
Left	39.3 (14)	61.1 (22)	19.4 (7)	80.6 (29)
Right	22.2 (8)	77.8 (28)	22.2 (8)	77.8 (28)
Elbow				
Left	27.8 (10)	72.2 (26)	44.4 (16)	55.6 (20)
Right	44.4 (16)	55.6 (20)	30.6 (11)	69.4 (25)
Knee				
Left	41.7 (15)	58.3 (21)	22.2 (8)	77.8 (28)
Right	47.2 (17)	52.8 (19)	19.4 (7)	80.6 (29)
Back	97.2 (35)	2.8 (1)	36.1 (13)	63.9 (23)

T=2.914,  $P=0.004$  were observed in comparison with controls.

### Multivariate analysis

Multivariate linear regression models for physical fitness, musculoskeletal complaints and psychological distress are presented in Fig. 1. All the necessary assumptions for multivariate linear regression were fulfilled. The  $R^2$  from all of the constructed models ranged from 0.134 to 0.598 (Fig. 1).

### Physical fitness

In the first model ( $R^2=0.598$ ), functional walking distance (6MWT) was independently negatively associated with BMI [B (s.e.)  $-6.8$  (2.3),  $P<0.0001$ ] and with GJH [B

(s.e.)  $-91.5$  (11.1),  $P<0.0001$ ]. Independent positive associations were found with dancers [B (s.e.) 75.9 (11.9),  $P<0.0001$ ]. No effect of PAL was found ( $P=0.862$ ).

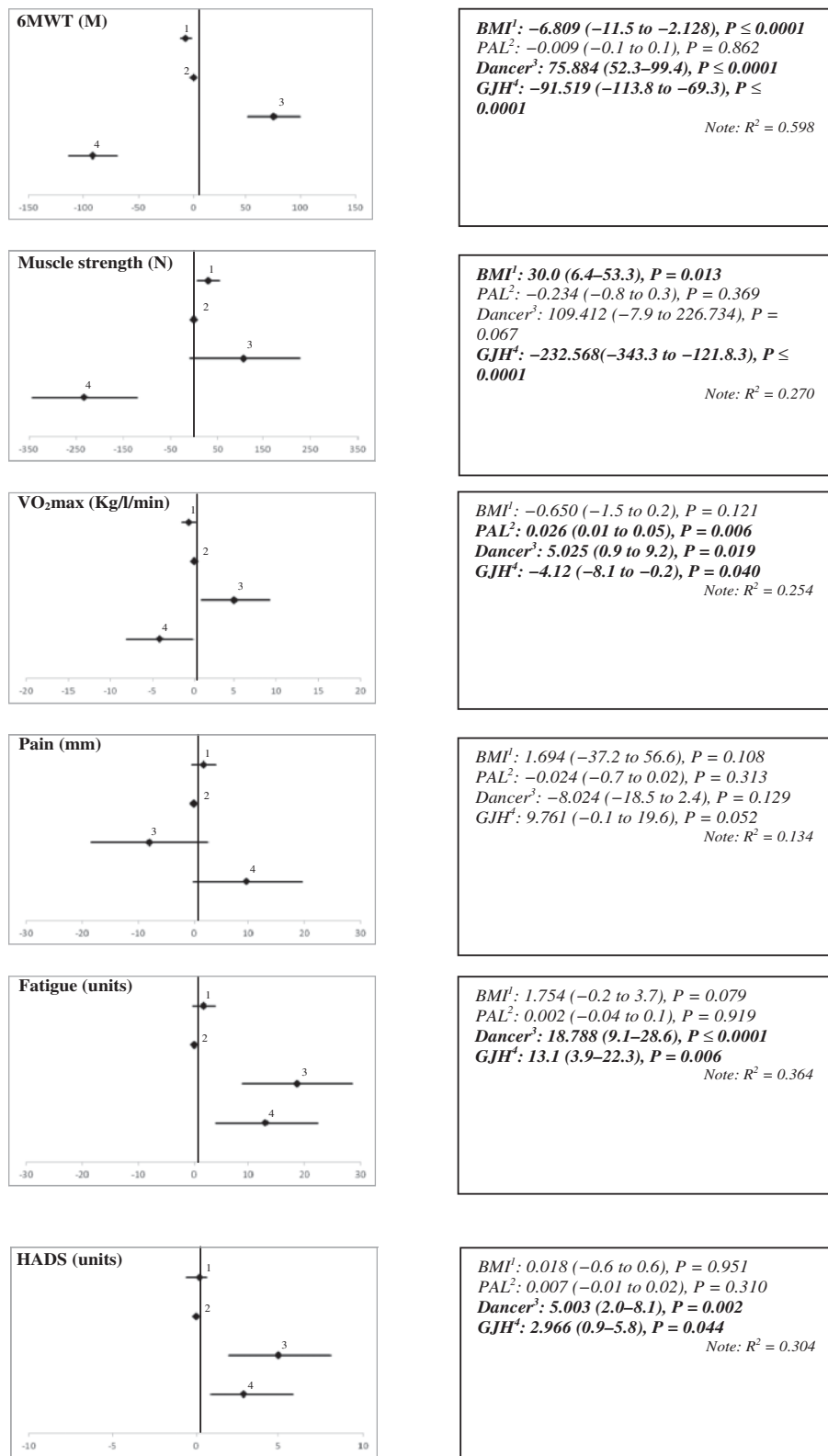
In the second model regarding muscle strength ( $R^2=0.270$ ), a significant positive association with BMI [B (s.e.) 30.0 (11.7),  $P=0.013$ ] was found. Muscle strength was negatively associated with the presence of GJH [B (s.e.)  $-232.6$  (55.5),  $P<0.0001$ ]. Both factors PAL and group (dancer/non-dancer) did not significantly contribute to the model. Although higher levels of muscle strength were found in dancers, this failed to reach significance [B (s.e.) 109.4 (58.8),  $P=0.067$ ].

In the third model ( $R^2=0.254$ ), submaximal exercise capacity was addressed. Positive associations between submaximal exercise capacity and the factor group [B (s.e.) 5.0 (2.1),  $P=0.019$ ] in favour of dancers were found. GJH was negatively associated with submaximal exercise capacity [B (s.e.)  $-4.1$  (2.0),  $P=0.040$ ]. This association was positively influenced by PAL [B (s.e.) 0.03,  $P=0.006$ ], whereas the factor BMI was not found to be significant ( $P=0.121$ ).

### Musculoskeletal complaints

When regarding pain ( $R^2=0.134$ ), no significant effects for any factor were found, although trends were found indicating that subjects with GJH had more severe pain [B (s.e.) 9.8 (4.9),  $P=0.052$ ] and dancers showed less intense pain [B (s.e.)  $-8.0$  (5.2),  $P=0.129$ ].

Fatigue ( $R^2=0.364$ ) was found to be significantly associated with group and GJH, indicating that subjects with GJH experience higher levels of fatigue [B (s.e.) 13.1 (4.6),  $P=0.006$ ], and dancers [B (s.e.) 18.8 (4.9),  $P<0.0001$ ] even more. The contribution of PAL and BMI to the model was found to be non-significant ( $P\geq 0.05$ ).

**Fig. 1** Linear regression factor analysis. Beta and corresponding 95% CI for all outcomes.



### Psychological distress

In the final models, psychological distress was addressed. Higher levels of total psychological distress ( $R^2=0.304$ ) were independently associated with dancers [B (s.e.) 5.0 (1.5),  $P=0.002$ ] and with GJH [B (s.e.) 3.0 (1.4),  $P=0.044$ ].

Similar results were found in the analysis of the subscales of the HADS in which higher scores on the anxiety subscale ( $R^2=0.288$ ) were found to be independently associated with dancers [B (s.e.) 2.9 (0.9),  $P=0.002$ ] and with GJH [B (s.e.) 1.8 (0.9),  $P=0.040$ ].

For the depression subscale ( $R^2=0.229$ ), higher scores were independently associated with dancers [B (s.e.) 2.1 (0.8),  $P=0.009$ ], but no significant association was found with GJH [B (s.e.) 1.1 (0.7),  $P=0.125$ ]. No effects of BMI and PAL were found ( $P \geq 0.05$ ).

### Discussion

The current study showed that even in more physically trained professional dancers, the presence of GJH is associated with lower muscle strength, lower submaximal exercise capacity and decreased functional walking distance. When regarding musculoskeletal complaints, the presence of GJH was associated with higher levels of fatigue and psychological complaints, especially with anxiety.

In the included subjects, GJH was significantly more present among dancers (66%) in comparison with control subjects (29%). The prevalence of GJH among control subjects was found to be high; however, it was within the range of values (0.6–31.5%) reported in the literature [3–5]. The prevalence within dancers was comparable with other studies (4.0–97%) [2]. Still, despite the higher prevalence of GJH among dancers, which was found to be a negative factor in physical fitness and functional walking distance, dancers performed better. And when comparing hypermobile dancers with hypermobile controls this difference was still present. This finding supports the assumption that enhancing physical fitness can be beneficial when regarding functional ability and motor competence [29]. Although we did not study the relationship of activity load with musculoskeletal complaints, the current data showed that overuse could potentially result in more frequent or more severe musculoskeletal complaints [30, 31]. Interestingly, dancers who follow a more physically challenging education experienced lower levels of pain intensity, again with GJH as a negative factor, in comparison with controls. This finding is in line with previous research that has demonstrated that muscle strength is associated with pain intensity in hypermobile individuals [32].

In contrast, fatigue levels were found to be higher in dancers and even more increased in hypermobile dancers. This could reflect the strenuous nature of the professional dance education, but also indicated that GJH was a negative modifier that could reflect additional strain because of the presence of more dynamic processes to maintain joint stability. When regarding the psychological distress, dancers were found to have higher

levels of depression and anxiety. Because of the decreased physical capabilities, in terms of muscle strength and stamina, it could be plausible that dancers with GJH have to make additional efforts to reach the requirements of professional dance education but also have additional physical challenges to maintain their skill level. This could result in additional psychological strain when considering the high physical demands associated with being a dancer. However, this is beyond the scope of the current study.

Evidence regarding the link between joint hypermobility and psychological complaints has been amassed in the past decade, indicating that psychological complaints like depression and anxiety are significantly more likely to be present in clinical [33] and non-clinical [34] populations with GJH. Still, although hypermobile dancers experienced more psychological distress than non-hypermobile dancers, both groups experienced significantly more psychological distress in comparison with controls. Although dancers were exposed to a more physical and potentially more psychologically strenuous educational programme, an alternative explanation could also be found at the level of genetic predisposition for the development of psychological complaints. Recent developments within the field of molecular psychiatry and neuropsychology have shown that genetic predispositions play an important role in behavioural patterns and the development of psychological disorders [35, 36]. From recently conducted research, it has been found that the clinical features of hypermobile joints, stretchy skin, arthralgia, fatigue and psychological complaints, often found in patients with hypermobility syndrome and Ehlers–Danlos syndrome hypermobile type, can potentially be linked to genetic deficiencies in tenascin-X [37, 38]. This evidence indicated a potential common genetic origin for disorders with GJH as a clinical feature [39–41]. When speculating, these commonalities in genetics and clinical presentation could also imply that genetic variations within the hypermobility phenotype could be a factor in the development of psychological complaints. In 2001 a common genetic vulnerability for anxiety disorders and hypermobility has been proposed, based on the report of a cytogenetic mutation (chromosome 15; identified as DUP25) within a family with a high incidence of panic disorders and hypermobility [42]. The latter finding could not be replicated in two other studies [43, 44]. This is beyond the scope of the current article; however, these questions could prove to be an interesting field for research and could prove to be valuable for clinical diagnostics, prevention and treatment [45].

The observed results should be interpreted in light of the following limitations: (i) the current study was limited to only females, and when considering the number of included variables this should be taken into account as a limitation to generalize these results to the normal population; (ii) the control group consisted of health care students who may take more care in personal health and may spend more time on issues of (personal) healthy living, which suggests that the difference may be larger when comparing with the general population.

Still, despite previously mentioned limitations, the present study showed indications that the presence of GJH is not as beneficial as assumed in the professional dance education. It is common practice to select students based on their joint mobility; however, caregivers should be aware that alongside the aesthetic advantages, a trade-off might exist in which hypermobile individuals are susceptible to the development of musculoskeletal and psychological complaints. This should not only be taken into account when selecting students to participate in professional dance education, but should also lead to more monitoring and awareness among caregivers and should lead to the development of effective (preventive) interventions for this category of patients.

### Conclusion

We conclude that GJH can also be viewed as a sign of vulnerability in terms of lower physical fitness, more musculoskeletal complaints and psychological distress in professional dancers. The pathological mechanism remains unknown, as does the reason why a mechanical factor like GJH is associated specifically with anxiety. This deserves further scientific exploration. Clinicians should be aware that individuals with GJH should be carefully monitored not only physically but also psychologically, especially within professional dance education.

#### Rheumatology key messages

- Even in trained dancers, GJH is a negative factor for physical and psychological functioning.
- Enhancing physical fitness could be beneficial in symptomatic GJH, but its effectiveness remains unproven.

### Acknowledgements

The authors would like to thank all the students from the Amsterdam School of Arts and the Amsterdam School of Health Professions for their participation and assistance during this study. The authors would also like to thank Margot Rijven, Menno de Vries for their efforts to realize this study.

**Funding:** This work was supported by the Amsterdam School of Health Profession, Education for Physical Therapy, Amsterdam, The Netherlands.

**Disclosure statement:** The authors have declared no conflicts of interest.

### References

- Grahame R, Jenkins JM. Joint hypermobility—asset or liability? A study of joint mobility in ballet dancers. *Ann Rheum Dis* 1972;31:109–11.
- Day H, Koutedakis Y, Wyon MA. Hypermobility and dance: a review. *Int J Sports Med* 2011;32:485–9.
- Larsson LG, Baum J, Mudholkar GS. Hypermobility: features and differential incidence between the sexes. *Arthritis Rheum* 1987;30:1426–30.
- Rikken-Bultman DG, Wellink L, van Dongen PW. Hypermobility in two Dutch school populations. *Eur J Obstet Gynecol Reprod Biol* 1997;73:189–92.
- Seckin U, Tur BS, Yilmaz O *et al.* The prevalence of joint hypermobility among high school students. *Rheumatol Int* 2005;25:260–3.
- Grahame R. Heritable disorders of connective tissue. *Baillieres Best Pract Res Clin Rheumatol* 2000;14:345–61.
- Malfait F, Hakim AJ, De Paepe A, Grahame R. The genetic basis of the joint hypermobility syndromes. *Rheumatology* 2006;45:502–7.
- Tinkle BT, Bird HA, Grahame R *et al.* 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 A* 2009;149A:2368–70.
- Hakim A, Grahame R. Joint hypermobility. *Best Pract Res Clin Rheumatol* 2003;17:989–1004.
- Rombaut L, Malfait F, De Wandele I *et al.* Muscle mass, muscle strength, functional performance, and physical impairment in women with the hypermobility type of Ehlers–Danlos syndrome. *Arthritis Care Res (Hoboken)* in press, doi: 10.1002/acr.21726.
- Grahame R, Hakim AJ. Hypermobility. *Curr Opin Rheumatol* 2008;20:106–10.
- Engelbert RH, van Bergen M, Henneken T, Helders PJ, Takken T. Exercise tolerance in children and adolescents with musculoskeletal pain in joint hypermobility and joint hypomobility syndrome. *Pediatrics* 2006;118:e690–6.
- Rombaut L, De Paepe A, Malfait F, Cools A, Calders P. Joint position sense and vibratory perception sense in patients with Ehlers–Danlos syndrome type III (hypermobility type). *Clin Rheumatol* 2009;29:289–95.
- Gazit Y, Nahir AM, Grahame R, Jacob G. Dysautonomia in the joint hypermobility syndrome. *Am J Med* 2003;115:33–40.
- Bulbena A, Gago J, Sperry L, Berge D. The relationship between frequency and intensity of fears and a collagen condition. *Depress Anxiety* 2006;23:412–17.
- Hincapié CA, Morton EJ, Cassidy JD. Musculoskeletal injuries and pain in dancers: a systematic review. *Arch Phys Med Rehabil* 2008;89:1819–29.
- Ostwald PF, Baron BC, Byl NM, Wilson FR. Performing arts medicine. *West J Med* 1994;160:48–52.
- 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;31:173–8.
- Wendel-Vos GC, Schuit AJ, Saris WH, Kromhout D. Reproducibility and relative validity of the short questionnaire to assess health-enhancing physical activity. *J Clin Epidemiol* 2003;56:1163–9.

- 20 Ainsworth BE, Haskell WL, Whitt MC *et al.* Compendium of physical activities: an update of activity codes and met intensities. *Med Sci Sports Exerc* 2000;32:S498–504.
- 21 Juul-Kristensen B, Rogind H, Jensen DV, Remvig L. Inter-examiner reproducibility of tests and criteria for generalized joint hypermobility and benign joint hypermobility syndrome. *Rheumatology* 2007;46:1835–41.
- 22 Atkinson HL, Nixon-Cave K. A tool for clinical reasoning and reflection using the international classification of functioning, disability and health (ICF) framework and patient management model. *Phys Ther* 2011;91:416–30.
- 23 Gulmans VA, van Veldhoven NH, de Meer K, Helders PJ. The six-minute walking test in children with cystic fibrosis: reliability and validity. *Pediatr Pulmonol* 1996;22:85–9.
- 24 Koblbauer IF, Lambrecht Y, van der Hulst ML *et al.* Reliability of maximal isometric knee strength testing with modified hand-held dynamometry in patients awaiting total knee arthroplasty: useful in research and individual patient settings? A reliability study. *BMC Musculoskelet Disord* 2011;12:249.
- 25 Keen EN, Sloan AW. Observations on the Harvard Step Test. *J Appl Physiol* 1958;13:241–3.
- 26 Macsween A. The reliability and validity of the Astrand nomogram and linear extrapolation for deriving  $\text{VO}_2\text{max}$  from submaximal exercise data. *J Sports Med Phys Fitness* 2001;41:312–17.
- 27 van de Putte EM, Engelbert RH, Kuis W, Kimpen JL, Uiterwaal CS. Alexithymia in adolescents with chronic fatigue syndrome. *J Psychosom Res* 2007;63:377–80.
- 28 Bjelland I, Dahl AA, Haug TT, Neckelmann D. The validity of the Hospital Anxiety and Depression Scale. An updated literature review. *J Psychosom Res* 2002;52:69–77.
- 29 Van Brussel M, Takken T, Uiterwaal CS *et al.* Physical training in children with osteogenesis imperfecta. *J Pediatr* 2008;152:111–16.
- 30 Russek LN. Hypermobility syndrome. *Phys Ther* 1999;79:591–9.
- 31 Kindermans HP, Roelofs J, Goossens ME *et al.* Activity patterns in chronic pain: underlying dimensions and associations with disability and depressed mood. *J Pain* 2011;12:1049–58.
- 32 Fatoye F, Palmer S, Macmillan F, Rowe P, van der Linden M. Pain intensity and quality of life perception in children with hypermobility syndrome. *Rheumatol Int* 2012;32:1277–84.
- 33 Ercolani M, Galvani M, Franchini C, Baracchini F, Chattat R. Benign joint hypermobility syndrome: psychological features and psychopathological symptoms in a sample pain-free at evaluation. *Percept Mot Skills* 2008;107:246–56.
- 34 Bulbena A, Agullo A, Pailhez G *et al.* Is joint hypermobility related to anxiety in a nonclinical population also? *Psychosomatics* 2004;45:432–7.
- 35 Cichon S, Craddock N, Daly M *et al.* Genomewide association studies: history, rationale, and prospects for psychiatric disorders. *Am J Psychiatry* 2009;166:540–56.
- 36 Stein MB, Stein DJ. Social anxiety disorder. *Lancet* 2008;371:1115–25.
- 37 Freitag S, Schachner M, Morellini F. Behavioral alterations in mice deficient for the extracellular matrix glycoprotein tenascin-R. *Behav Brain Res* 2003;145:189–207.
- 38 Kawakami K, Matsumoto K. Behavioral alterations in mice lacking the gene for tenascin-X. *Biol Pharm Bull* 2011;34:590–3.
- 39 Voermans NC, Altenburg TM, Hamel BC, de Haan A, van Engelen BG. Reduced quantitative muscle function in tenascin-X deficient Ehlers–Danlos patients. *Neuromuscul Disord* 2007;17:597–602.
- 40 Voermans NC, Bonnemann CG, Huijings PA *et al.* Clinical and molecular overlap between myopathies and inherited connective tissue diseases. *Neuromuscul Disord* 2008;18:843–56.
- 41 Voermans NC, Jenniskens GJ, Hamel BC *et al.* Ehlers–Danlos syndrome due to tenascin-X deficiency: muscle weakness and contractures support overlap with collagen VI myopathies. *Am J Med Genet A* 2007;143A:2215–19.
- 42 Gratacos M, Nadal M, Martin-Santos R *et al.* A polymorphic genomic duplication on human chromosome 15 is a susceptibility factor for panic and phobic disorders. *Cell* 2001;106:367–79.
- 43 Tabiner M, Youings S, Dennis N *et al.* Failure to find DUP25 in patients with anxiety disorders, in control individuals, or in previously reported positive control cell lines. *Am J Hum Genet* 2003;72:535–8.
- 44 Weiland Y, Kraus J, Speicher MR. A multicolor fish assay does not detect DUP25 in control individuals or in reported positive control cells. *Am J Hum Genet* 2003;72:1349–52.
- 45 Remvig L, Engelbert RH, Berglund B *et al.* Need for a consensus on the methods by which to measure joint mobility and the definition of norms for hypermobility that reflect age, gender and ethnic-dependent variation: is revision of criteria for joint hypermobility syndrome and Ehlers–Danlos syndrome hypermobility type indicated? *Rheumatology* 2011;50:1169–71.