

Functional ability and physical and psychosocial well-being of hypermobile schoolchildren

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ABSTRACT

Objective. To compare the functional ability and the physical and psychosocial well-being of children with joint hypermobility to those of age- and sex-matched non-hypermobile subjects.

Methods. 311 healthy Italian school-children aged 6.3 to 19.3 years were examined for hypermobility of the joints. Functional ability was assessed through the Childhood Health Assessment Questionnaire (CHAQ) and the physical and psychosocial well-being through the Childhood Health Questionnaire (CHQ). The parent's assessment of the child's overall well-being and of the child's pain was measured on a visual analogue scale.

Results. The overall prevalence of articular hypermobility was 34% (106/311), with the median hypermobility score being 3 (interquartile range 1, 5). Although the hypermobility score of girls (median 3; interquartile range 2, 5) exceeded that of boys (median 2.5; interquartile range 0, 5), this difference was not statistically significant ($p = 0.16$). The level of hypermobile children's pain in the preceding weeks, as assessed by the parents, was comparable to that recorded in the non-hypermobile peers. There was a weak negative correlation between the hypermobility score and the age of the child ($r = -0.14$, $p = 0.01$). All instrument scores were comparable between hypermobile and non-hypermobile subjects, with the sole exception of a borderline significant greater impairment of the Role/social limitations-physical subscale of the CHQ in the hypermobile group. The hypermobility score was not correlated with any instrument score.

Conclusions. The presence of joint hypermobility does not affect the functional ability and the physical and psychosocial well-being of otherwise healthy children. These results suggest that the physical functioning in everyday life and the general health status of hypermobile children are not impaired.

Introduction

Joint hypermobility is detectable in a substantial proportion of healthy children and is affected by many factors, including age, sex, and probably, race

(1). Several studies have suggested that this condition may be associated with musculoskeletal complaints, predominantly pain (so-called benign joint hypermobility syndrome, BJHS) (2-5), although others have not found such a link (6,7). It has been suggested that for the most affected children the recurrence of pain symptoms may limit not only the physical or sporting activities but also the everyday activities of life (8, 9). However, the impact of joint hypermobility on children's physical functioning and general health status has not been well documented.

The purpose of this study was to compare the functional ability and the physical and psychosocial well-being of children with joint hypermobility to those of non-hypermobile peers.

Methods

Three hundred and eleven healthy Italian children (156 boys and 155 girls) aged 6.3 to 19.3 years (median 10.6 years; interquartile range 8.5, 14.1 years) attending school in Pavia, Italy were examined for joint hypermobility. The age distribution of the subjects was the following: 134 (43.1%) were 6.3 to 10 years of age; 82 (26.4%) were 10.1 to 13 years old; and 95 (30.5%) were 13.1 to 19.3 years old. The subject sample was representative of children attending the primary and secondary schools in the study area. No child was found to have the features of a heritable disorder of connective tissue disease, such as Ehlers-Danlos syndrome, Marfan syndrome, or osteogenesis imperfecta. The non-mechanical scoring system described by Carter and Wilkinson (10) with the modification of Beighton *et al.* (11) was used. This method is based on 5 manoeuvres, 4 of which are performed bilaterally. The maneuvers used in this scoring system are: (1) extension of the wrist and metacarpal phalanges so that the fingers are parallel to the dorsum of the forearm (1 point for each hand) – 2 points; (2) passive apposition of the thumbs to the flexor aspects of the forearm (1 point for each thumb) – 2 points; (3) hyperextension of the elbows beyond 10° (1 point for each elbow) – 2 points; (4) hyperextension of the knee beyond a 10° (1 point

for each knee) – 2 points; and (5) forward flexion of the trunk with knees fully extended so that the palms of the hands rest flat on the floor – 1 point. Possible scores range from 0 to 9. In our study, a score of 5 or greater was considered sufficient to classify the child as hypermobile. Two examiners (CM and MB) together assessed all subjects for hypermobility. Both had to agree that a joint was hypermobile for it to be graded as such. Informed consent for study participation was obtained from the parents of all children.

After the joint mobility assessment, one parent of each child was asked to complete the Italian version of the Childhood Health Assessment Questionnaire (CHAQ), an instrument designed to measure functional ability in children (12), and the Italian version of the Child Health Questionnaire (CHQ) (12), an instrument designed to measure the physical and psychosocial well-being of children. Briefly, the CHAQ measures the child's ability to perform functions in 8 areas (Dressing and Grooming, Arising, Eating, Walking, Hygiene, Reaching, Gripping, and Activities). The scores of the 8 functional areas are averaged to calculate the CHAQ disability index (DI), which ranges from 0 to 3 (0=best; 3=worst). The CHAQ includes the parent's assessment of the child's overall well-being and the child's pain, both on a 10-cm visual analogue scale (VAS) (0=best; 10=worst). The CHQ is a 50-item, parent-completed questionnaire. It measures components of physical and psychosocial functioning on 15 subscales. The scores in each subscale range from 0 to 100, with the higher score indicating better well-being. These subscales contribute to generate 2 summary scores: a physical summary score (PhS) and a psychosocial summary score (PsS), which have a mean of 50 ± 10 in the general European population (13).

Statistical analysis

The comparison of demographic features and instrument scores between hypermobile and non-hypermobile children was made by the Mann-Whitney U test. The relationship between the

hypermobility score and the demographic features and instrument scores was evaluated through the Spearman rank correlation method. Multiple logistic regression was used to find independent variables associated with the presence of hypermobility (Beighton score ≥ 5). Before the application of logistic regression procedures, some variables were dichotomized to binary variables. Cut-off points for the children's age were < 11 years and ≥ 11 years; cut-off points for the CHAQ score and the parent's global assessment of the child's overall well-being and of the child's pain were 0 and > 0 ; cut-off points for the CHQ-PhS, the CHQ-PsS and the CHAQ subscales were < 50 and ≥ 50 .

The step-down strategy of analysis was chosen; it consists of examining the effect of removing variables from the saturated model. Variables that were considered important *a priori* for the outcome or were statistically significant in the univariate analysis ($p < 0.05$) were entered in the model. The variables analysed were the following: gender, age, CHAQ score, parent's global assessment of the child's overall well-being and the child's pain, CHQ-PhS, CHQ-PsS, and the CHQ subscales Role/social limitations–emotional behavioral and Role/social limitations–physical. The effect was expressed in terms of the odd ratio (OR) and 95% confidence interval (CI) and the statistical significance was tested by means of the likelihood ratio test (LR test). The statistical package used were the "Statistica" (StatSoft Corp., Tulsa, OK) for univariate analyses and the "Stata release 7" (Stata Corporation, Texas, USA) for multivariate analyses.

Results

The overall prevalence of articular hypermobility in our healthy schoolchildren was 34% (106/311), with the median hypermobility score being 3 (interquartile range: 1, 5). The median Beighton score was 6 (interquartile range 5, 6) in hypermobile children and 2 (interquartile range 0, 3) in non-hypermobile children ($p < 0.0001$). Although the hypermobility score of girls (median 3; interquartile range 2, 5) exceeded that

of boys (median 2.5; interquartile range 0, 5), this difference was not statistically significant ($p = 0.16$). There was a weak negative correlation between the hypermobility score and the age of the child ($r = -0.14$, $p = 0.01$).

Tables I and II show the comparison of the CHAQ and CHQ scores between non-hypermobile and hypermobile subjects and the correlation between the same instrument scores and the hypermobility score. All instrument scores were comparable between hypermobile and non-hypermobile subjects, with the sole exception of a borderline significant greater impairment of the Role/social limitations–physical subscale of the CHQ in the hypermobile group. The hypermobility score was not correlated with any instrument score.

The logistic regression analysis showed that the only variable that entered the best fitting model corrected for the children's age was the parent's global assessment of the child's overall well-being ($= 0 / > 0$), which was associated with the absence/presence of hypermobility with an OR of 1.73 (95% CI: 1.02–2.91; $p = 0.04$).

Discussion

We found joint hypermobility in 34% of Italian normal schoolchildren. This prevalence is in the highest range of that previously reported in populations of schoolchildren in different countries, which varies from 5% to 34% (7, 14–18). The discrepancy among studies may be attributable to age and racial variation of the children's sample, differences in the scoring system used, and/or examiners' bias.

It is important to distinguish hypermobility, which describes the often asymptomatic increased range of motion of joints, from hypermobility syndrome, its symptomatic counterpart. Indeed, in the recent years it has been suggested that joint hypermobility may have a relevant impact on individual's lives, due to its frequent association with musculoskeletal symptoms or signs, particularly pain (8, 9). For this association, the term BJHS has been proposed, together with a set of diagnostic criteria, the so-called Brighton criteria (2). However, the Brighton criteria

Table I. Comparison of the parent's global assessments of the child's overall well-being, the parent's assessment of the child's pain, the CHAQ disability index, and the scores of the 8 functional areas of the CHAQ between non-hypermobile and hypermobile children and correlation of the same instrument scores with the hypermobility score.

	Non-hypermobile children			Hypermobile children			Plevel [§]	Correl. with HM score	
	No	Median	(IQR)	No	Median	(IQR)		R [£]	Plevel [£]
Patients' age (years)	205	10.9	(9, 14.2)	106	10	(8.1, 13.8)	0.12	-0.14	0.01
Hypermobility score [¶]	205	2	(0, 3)	106	6	(5, 6)	< 0.0001	-	-
Parent's global assessment [#]	190	0	(0, 0.2)	93	0.1	(0, 0.2)	0.05	0.08	0.21
Parent's pain assessment [#]	188	0	(0, 0.1)	90	0	(0, 0.1)	0.10	0.06	0.32
CHAQ disability index [§]	205	0	(0, 0.3)	106	0.1	(0, 0.3)	0.11	0.11	0.05
Dressing and grooming [§]	205	0	(0, 1)	106	0	(0, 1)	0.28	0.09	0.12
Arising [§]	204	0	(0, 0)	106	0	(0, 0)	0.15	-0.09	0.12
Eating [§]	204	0	(0, 0)	106	0	(0, 0)	0.54	0.08	0.17
Walking [§]	204	0	(0, 0)	106	0	(0, 0)	0.31	0.00	0.96
Hygiene [§]	204	0	(0, 0)	106	0	(0, 0)	0.21	0.09	0.10
Reaching [§]	204	0	(0, 0)	106	0	(0, 0)	0.60	0.05	0.36
Gripping [§]	204	0	(0, 0)	106	0	(0, 0)	0.42	0.10	0.09
Activities [§]	204	0	(0, 1)	106	0	(0, 1)	0.26	0.08	0.16

IQR: interquartile range; CHAQ: Childhood Health Assessment Questionnaire; [§]Mann-Whitney U test test; [£]Spearman rank correlation coefficient; HM: hypermobility; [¶]on a 0-9 scale (0=normal; 9=greatest hypermobility); [#]on a 10-cm visual analogue scale (0=best; 10=worst); [§]on a 0-3 scale (0=best; 3=worst).

Table II. Comparison of the CHQ summary scores and CHQ subscales between non-hypermobile and hypermobile children and correlation of the same instrument scores with the hypermobility score.

	Non-hypermobile children			Hypermobile children			Plevel [§]	Correlation with HM score [¶]	
	No	Median	(IQR)	No	Median	(IQR)		R [£]	Plevel [£]
Physical summary score [§]	190	55.3	(53.6, 56.5)	97	54.7	(53, 56.4)	0.33	-0.03	0.63
Psychosocial summary score [§]	190	51.9	(46.3, 55.9)	97	52.8	(47, 57.7)	0.19	0.06	0.28
Global health [#]	199	85	(85, 100)	105	85	(60, 100)	0.27	-0.05	0.42
Physical functioning [#]	203	100	(100, 100)	105	100	(100, 100)	0.85	0.01	0.89
Role/social limitations- emotional behavioral [#]	202	100	(100, 100)	104	100	(100, 100)	0.04	0.11	0.06
Role/social limitations-physical [#]	203	100	(100, 100)	106	100	(100, 100)	0.04	-0.09	0.13
Bodily pain/discomfort [#]	203	90	(70, 100)	105	100	(70, 100)	0.81	0.01	0.83
Behavior [#]	200	80.8	(72.5, 89.2)	100	80.8	(72.5, 91.7)	0.69	0.02	0.76
Global behavior [#]	200	85	(60, 85)	103	85	(60, 100)	0.35	-0.06	0.32
Mental health [#]	202	75	(65, 85)	102	77.5	(65, 85)	0.52	0.01	0.85
Self esteem [#]	202	79.2	(70.8, 90)	102	83.3	(75, 91.7)	0.10	0.09	0.12
General health perceptions [#]	203	80.8	(68.3, 87.5)	104	79.2	(68.3, 87.5)	0.50	0.01	0.91
Change in health [#]	203	50	(50, 75)	105	50	(50, 75)	0.41	0.03	0.62
Parent impact-emotional [#]	201	83.3	(58.3, 91.7)	104	75	(58.3, 91.7)	0.77	-0.04	0.44
Parent impact-time [#]	200	100	(88.9, 100)	102	100	(88.9, 100)	0.91	-0.03	0.66
Family activities [#]	200	100	(87.5, 100)	104	100	(87.5, 100)	0.48	0.02	0.68
Family cohesion [#]	201	85	(60, 85)	106	85	(60, 85)	0.75	0.01	0.82

IQR: interquartile range; CHQ: Child Health Questionnaire; [§]Mann-Whitney U test test; [£]Spearman rank correlation coefficient; HM: hypermobility; [¶]The physical and psychosocial summary scores have a mean of 50 ± 10 in the general European pediatric population; [#]the scores in each CHQ subscale range from 0 to 100, with the higher score indicating better well being; [¶]on a 0-9 scale (0=normal; 9=greatest hypermobility).

have not been yet validated in children under 16 years of age, which is the age range of most of the subjects included in our study. Recently, Engelbert *et al.* (19) found that clinically manifest

symptoms in otherwise healthy children with generalized joint hypermobility are accompanied by increases in the laxity of other body tissues, suggesting that generalized joint hypermo-

bility with musculoskeletal symptoms is not restricted to joint tissues. The fact that in symptomatic hypermobile children a more systemic derangement was also present as compared with

asymptomatic hypermobile children were consistent with a natural interindividual variation in connective tissue composition as an explanation for different degrees of clinical symptomatology.

We found that the presence of joint hypermobility did not affect functional ability or physical and psychosocial well-being of healthy children, although the multivariate analysis showed that parents of hypermobile children tended to judge as worse their children's overall well-being on a VAS as compared to parents of non-hypermobile children. Although we did not assess specifically the occurrence of musculoskeletal pain in the life of hypermobile children, the degree of their pain in the preceding weeks measured by their parents either on a VAS or through the Bodily pain/discomfort subscale of the CHQ was comparable to that recorded in non-hypermobile peers. Taken together, these findings suggest that joint hypermobility does not affect significantly the physical functioning and the physical and psychosocial well-being of healthy children.

Our study should be viewed in the light of certain limitations. Although the Beighton 9-point scoring system is widely used as a clinical screening test for hypermobility, it cannot be relied upon to identify pauciarticular hypermobility, where a hypermobile joint fall outside the five areas sampled in the score (20). Because parents rather than the children themselves completed the health status questionnaires, it is possible that the health perceptions of the children may have differed from those of the parents. Furthermore, the instruments we used were developed to

assess health status in children with chronic diseases and may not have enough power to detect smaller differences from a normal population sample. In addition, they do not include the evaluation of the ability of children to perform more demanding physical or sport activities, which are more likely to be affected by joint hypermobility. Concerning the CHAQ and the enclosed VAS, an additional potential shortcoming is the fact that they only measure symptoms over the previous week. Because symptoms and problems related to hypermobility and to associated mechanical musculoskeletal conditions might be intermittent and children may have periods without symptoms, a 1-week period can be too brief to capture all potential complaints.

We conclude that the presence of joint hypermobility does not affect the functional ability and the physical and psychosocial well-being of otherwise healthy children. These results suggest that the physical functioning in everyday life and the general health status of hypermobile children are not impaired.

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