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Joint Hypermobility Classes in 9-year-old Children from the General Population and Anxiety Symptoms

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Abstract

Objective: To obtain joint hypermobility classes in children from the general population and to study their characteristics in relation to anxiety measures.

Methods: 336 9-year-old children from the general population were clinically assessed through 9 items of hypermobility and their parents reported about the severity of anxiety symptoms. Latent class analysis (LCA) was estimated to group the children according to the presence of hypermobility symptoms and the obtained classes were related to anxiety.

Results: A 2-class solution, labeled as high hypermobility and low hypermobility, best fitted the data. Children in the high hypermobility group presented higher scores in separation anxiety, social phobia, physical injury fears and total anxiety scores than those in the low group. When applying the threshold reference scores to the total anxiety score, a 7.4% of children in the high hypermobility group versus 6% in the low group were reported to experience clinical elevations on total anxiety.

Conclusions: High symptoms of hypermobility are associated with higher scores in anxiety symptoms in children from the general population. Children with frequent symptoms of hypermobility may benefit from screening for anxiety symptoms because a subset of them are experiencing clinical elevations and may be necessary to provide them with comprehensive physical and psychological treatment.

Key-words: Anxiety; Children; Joint hypermobility syndrome

Joint Hypermobility Classes in 9-year-old Children from the General Population and Anxiety Symptoms

Joint hypermobility syndrome (JHS) is a connective tissue hereditary disorder characterized by increased distensibility of the joints in passive movements and hypermobility in active movements, which is not explained by other rheumatologic disease (1). JHS is quite prevalent in the adult general population (10-20%; female-male ratio 3:1), although it is frequently undiagnosed (2). The classification and definition of JHS is still under debate. The 2017 International Classification of the Ehlers-Danlos Syndromes (EDS) (3) includes the category Hypermobile EDS (hEDS) for unresolved forms of EDS III and hypermobility, and defines Generalized Joint Hypermobility (GJH) as one of the major criteria together with skin hyperextensibility and atrophic scarring, indicating that it should be assessed according to the Beighton score (a score \geq 5 is considered positive for GJH).

In adults, the condition is associated with articular pain, recurrent dislocations, scoliosis, fibromyalgia, hernias, ecchymoses, hypertrophic scars, rectal, uterine and mitral valve prolapses, pneumothorax, dysautonomia, and thyroid dysfunction (4). Glycoprotein deficiency and genetic alterations affecting collagen formation have been proposed as the mechanisms that explain the tissue looseness (4). However, JHS has not only been associated with physical and somatic symptoms, but also with emotional symptomatology. Specifically, individuals with JHS are at an increased risk of anxiety disorders. Review studies have reported a high prevalence of anxiety disorders, particularly panic disorder (PD) and agoraphobia (A), greater perception of fear and greater anxiety symptoms in individuals with JHS in comparison with those without (5-7). The underlying mechanisms behind this association include genetic risks, increased

exteroceptive and interoceptive mechanisms and decreased proprioception (8, 9). Both cross-sectional and longitudinal studies have reported this association together with a high degree of heritability, pointing towards an underlying genetic mechanism. On the other hand, dysautonomia and increased interoceptive perception are shared features of both anxiety and JHS), indicating that key neural mechanisms may also be involved (10). Neuroimaging studies in young adults have also shown an increased response in emotion processing brain areas, which could explain the high affective reactivity seen in JHS (10). There is also evidence for atypical regulation of the autonomic nervous system (10, 11). New clinical phenotypes have more recently been put forward to provide comprehensive models that include different somatic, psychological, cognitive and behavioral aspects involved in this multidimensional condition (9).

Most of the literature about the association between JHS and anxiety focuses on adults. JHS, however, is common in childhood and tends to decline with age (12, 13). It has been identified in the general population of different cultures with prevalences ranging from 7% to 54.1% for ages 3 to 6 (14, 15) and from 5.7% to 39.3% for ages 10 to 18 (16, 17, 18). These discrepancies in prevalence may be explained, in part, by the different methodologies used, such as the lack of consensus regarding the cut-offs used to define JHS and the different growth stages of the samples studied, highlighting the need for further studies on the definition of JHS in children (18). Like in adults, JHS in children has been associated with several physical complications, such as multi-systemic complaints (19), poor quality of life and disabling fatigue (20).

Notwithstanding, there are no studies on the concomitant emotional disorders and symptoms of JHS in children.

Anxiety disorders together with behavior disorders are the most prevalent mental disorders among children (21). In Western industrialized countries the prevalence of

anxiety disorders is similar (lifetime prevalence approximately 15-20%), whereas for Asian and African American children it is lower (22). In Spanish children from the general population the prevalence of anxiety disorders is 11.8% (23) and for 6-9 year-old outpatients it is 13.3% (24). The prevalence of some anxiety disorders, such as agoraphobia and panic, generalized anxiety, and obsessive compulsive disorders tends to increase with age and development, while the prevalence of others tends to decrease, as is the case with separation anxiety and specific phobias (22). Once anxiety disorders start, they usually have a recurrent or chronic course, causing individuals substantial suffering. This is also true for levels of anxiety under the diagnostic threshold. In this line, Bel-Dolan et al. (25) found that children with subclinical anxiety, in comparison with nonanxious, endorsed higher levels of anxiety, depression and loneliness. For this reason, detecting anxiety early is important, as permits to identify high-anxious children, to prevent anxiety or to intervene closer to onset (26, 27). Identifying at-risk groups, like children with hypermobility as in the present work, helps to focus preventive efforts. Consequently, the aim of this study is to obtain classes of JHS in children from the general population and to study the characteristics of this syndrome in relation to anxiety measures. We opted for a person-centered approach that groups individuals according to the characteristics of different features and focuses attention on the intra-individual structure of variables, with the advantage that the children are conceived as a whole rather than the sum of isolated features (28, 29). Profiles obtained with this methodology are well suited for addressing questions concerning group differences in patterns of clinical profiles. In line with the results for adult in the literature, we expect children pertaining to classes with high hypermobility to present higher anxiety symptomatology in comparison with children pertaining to classes with low hypermobility. Like physical therapy, which plays a central role in the management

of the physical symptoms of JHS (30), psychological therapy may also be required to deal with the emotional symptoms if also present.

Materials and Methods

Participants

The data are part of a large-scale longitudinal study of behaviour problems in children from age 3, who were screened for behaviour problems and followed up annually until age 9 (the design procedure is detailed in (31)). The two-phase design involved selecting a random sample of 2,283 children from the census of in-school 3-year-old preschoolers of a city in Spain. A total of 1,341 families (58.7%) agreed to participate in the first phase of the study and there were no sex differences ($p=.95$) between those who agreed to participate and those who declined. Participation, however, was greater among high socioeconomic (SES) families than low-status families ($p<.001$). The screening for children in the second phase was carried out using the conduct problem scale of the Strengths and Difficulties Questionnaire (SDQ³⁻⁴) (32). A random sample of 30% of the children with a negative screening score and all the children with a positive score were invited to remain as part of the longitudinal research process. The second-phase sample included 622 families (10.6% of those invited declined to participate in the second phase). There were no differences in SDQ emotional problems mean scores for the children between the first and second phases of the design ($p= .382$). No differences were found on comparing participants and refusals by sex ($p=.82$) or by type of school ($p=.85$).

At age 9, which corresponds to the seventh follow-up, 455 (73.2%) children remained in the study and all the data analysed in the present research were available for 336 of these children (174 boys; 51.9%). By focusing on a sample group of 9-year-

olds, an age at which neither sex is expected to have yet reached puberty, the possible effect of hormones on both anxiety and JHS are controlled for (33, 34). Socioeconomic level was distributed as follows: 41.2% high, 46.9% mean-high/mean and 11.9% mean-low/low. The ethnic composition of the sample is representative of the population at large: 93.9% of the children were Caucasian, 3.1% were Hispanic-American, 1.1% were oriental and 1.9% pertained to other ethnic groups. There were no statistical differences in the variables of the study by socioeconomic level ($p = .179$ for anxiety and $p = .991$ for hypermobility).

Instruments

Joint hypermobility. Before starting the study, the examiners took a training course on all Beighton's and Hospital del Mar items. Kappa values against the gold standard were obtained and retraining was offered if performance was poor. As expected, correlation between the two systems was high (near 0.9), and so the most reliable items together with the most feasible ones in terms of the characteristics of the sample to be examined, were selected. Joint hypermobility was assessed through 9 items that included all the indicators of Beighton (1) (items 1, 2, 3, 6, 9) and some of the indicators of the Hospital del Mar (35) (items 4, 5, 7) that were appropriate for 9 year-old children, plus an additional item of head rotation (item 8) (see Table 1). Head rotation was included not only because it is very often considered by clinical rheumatologists, but also because it is increasingly examined by neurologists in relation to functional conditions (36). The hypermobility schedule defines the criteria to consider if hypermobility is present or absent for each item and shows a picture to illustrate the item. Evaluators scored each item through clinical maneuvers of the child's joints and the right and left sides of the body were assessed for all measurements,

except for head and trunk. There were no differences in the distribution of the presence of hypermobility for the items between the left and right sides. Hypermobility was therefore considered as present if there was hypermobility on either of the two sides. The 9 items, scored dichotomously (1=*present*; 0=*absent*), were summed up to calculate a total score. Inter-rater agreement between each evaluator and the senior author (A.B.) ranged from kappa values of .65 to .94.

The *Spence Children's Anxiety Scale-Parent version (SCAS-P)* (37) assesses the severity of anxiety symptoms following the anxiety disorder categories proposed by the DSM-IV. The parent version contains 38 items (0:*never* to 3:*always*) that assess six domains of anxiety. Although there are not defined cut-off scores for clinical anxiety, the author suggested that T scores above 60 as indicative of sub-clinical or elevated levels of anxiety (38). The 6 subscales are (between brackets number of items, ordinal alpha (39), cut-off score for girls/boys): separation anxiety (6 items, $\alpha_o = .73$, 7/7), social phobia (6 items, $\alpha_o = .79$, 7/7), obsessive compulsive disorder (6 items, $\alpha_o = .83$, 3/3), panic/agoraphobia (9 items, $\alpha_o = .88$, 2/2), physical injury fears (5 items, $\alpha_o = .66$, 6/5) and generalized anxiety (6 items, $\alpha_o = .78$, 5/5). A total score indicates the severity of the anxiety symptomatology (38 items, $\alpha_o = .92$, 28/26). Raw scores are obtained summing the answers to the items.

Procedure

The longitudinal project was approved by the ethics review committee of the authors' institution (Ethics Committee for Human and Animal Experimentation). Informed written consent was obtained from the parents of the children participating in the study. The families were selected for participation and assessed at the schools. Twelve graduated psychologists were trained in hypermobility assessment, during

which they had to obtain good agreement ($\kappa > .61$) (40) with the criterion assessment (senior author) in 12 cases to be ready to do the assessment. Each child was assessed individually.

Statistical analysis

First, the percentage of presence for each hypermobility item between boys and girls was compared through exact test for comparison of proportions. Student's *t*-test was used for sex comparison of the total hypermobility score, the six SCAS-P dimensions and the total anxiety score. To improve the description of anxiety measures, percentiles 5 and 95 were calculated for the whole sample and separately by sex.

Latent class analysis (LCA) was estimated to group children based on presence of hypermobility in the 9 items. LCAs with one to five classes were conducted using a robust maximum likelihood estimator (MLR). Because the multistage sampling gave unequal probabilities of being selected depending on the screening group, analyses were weighted by the inverse proportion to the probability of participants' being selected in the second phase of the project. To determine the optimum number of classes, several goodness of fit indexes were used (see ahead) plus the best clinical interpretability. Proportion comparison tests between the groups finally selected were then estimated for each hypermobility item and Student's *t*-test for the total hypermobility score.

Comparison of quantitative anxiety measures and total hypermobility score between the classes obtained was done using Student's *t*-test and Cohens' *d* effect size. Cohens' *d* effect size was considered small for values $|d| \leq 0.2$, medium for $|d| \approx 0.5$ and large for $|d| \geq 0.8$ (41). Finally, percentage of presence for each hypermobility item and percentage of high anxiety for the total and the 6 subscales were compared between classes using chi-square test and risk ratio.

LCA was conducted with Mplus7.11 and the rest of the analyses were conducted with Stata 14. The type-I error was set at the usual .05. No correction of the alpha familywise error rate was applied because the exploratory nature of the work suggested the rate of false negatives should be minimized and Bonferroni's or similar correction procedures could increase this rate (42).

Results

Hypermobility and anxiety: Description and comparison by sex

As shown in Table 2, hypermobility was present for more than half of the sample in 7 of the 9 items, that is, all of them except knee hyperextension (35.9%) and trunk (39.3%). Knee hyperflexion (81.8%), fifth finger (75.6%), shoulder (74.0%) and patella (73.4%) were the most prevalent. The total hypermobility score showed a high mean value of 5.67 on a range 0-9. When comparing hypermobility between sexes, the only difference found was in trunk ($p<.001$), with girls (51.5%) being more flexible than boys (27.9%).

Table 2 also presents descriptives for anxiety scores. Considering the number of items summed up for each scale, social phobia and separation anxiety obtained the highest average, while panic/agoraphobia and obsessive compulsive obtained the lowest. The only sex difference was in physical injury fears ($p=.035$), with girls showing higher scores.

In the absence of statistical differences between sexes, the rest of the analyses were conducted for the whole sample.

Latent classes of hypermobility

Table 3 shows goodness of fit indexes for the LCA analysis carried out with one to five classes. The solution with two classes was selected based on better

interpretability and on the five abovementioned statistical criteria (43): (1) larger decrement in AIC ($AIC_{2c} = 3520.9$ vs $AIC_{1c} = 3621.9$), (2) lowest BIC ($BIC_{2c} = 3593.0$), (3) the adjusted Lo-Mendell-Rubin likelihood ratio test, which gave non-significant p -values for 3, 4 and 5 classes solutions, suggesting that a model with one fewer class (2 classes) is preferable, (4) groups with more than 5% of the total sample ($n_1 = 135, 40.2\%$ and $n_2 = 201, 59.8\%$) and (5) posterior probabilities of belonging to the assigned class between .84 and .94 (above 0.70, as recommended). The selected solution only had worse entropy (.644) than other solutions (.855 for the 4-class and .845 for the 5-class models).

With the aim of characterizing the two selected groups with respect to its hypermobility, the comparison of the 9 items and the total score is shown at the top of Table 4. Group 1 scored statistically lower than group 2 in seven items and the difference in Trunk was almost significant ($p=.055$). Only the most prevalent item, knee hyperflexion, was endorsed similarly in the two groups ($p=.761$). The groups also differed greatly in the total hypermobility score ($p<.001, d=2.29$). These results allowed us to label groups as ‘low hypermobility’ (group 1) and ‘high hypermobility’ (group 2).

Association of the hypermobility groups with anxiety

The bottom part of Table 4 shows the comparisons for anxiety measures between the high and low hypermobility classes, as well as percentiles 5 and 95 in each class. Statistically significant differences were found for separation anxiety ($p=.007$), social phobia ($p=.010$), physical injury fears ($p=.013$) and the total score ($p=.003$), with children in the high hypermobility group being assessed as more anxious by their parents. The difference for generalized anxiety disorder was almost significant ($p=.053$). Effect sizes for significant comparisons were small to moderate.

Table 1 online shows percentages of high anxiety obtained after applying threshold scores to quantitative measures for the whole sample and the two hypermobility groups. Globally, the percentage of children in the high anxiety ranges from a 4.2% for obsessive compulsive to 15.5% for generalized anxiety. Comparison between groups revealed statistically significant differences for social phobia ($p=.050$) and physical injury fears ($p=.048$). All risk ratio except for generalized anxiety are greater than one, reflecting more clinical anxiety in high hypermobility group.

Discussion

We obtained two classes of JHS symptoms in children from the general population, labeled as high and low hypermobility, which showed differences in patterns of clinical profiles of anxiety. As reported in the literature on adults with JHS, children with high symptomatology of hypermobility showed more severe anxiety symptomatology than those with low hypermobility symptoms. The results confirm the heterogeneity of the presentation of JHS, a syndrome still quite unrecognized among professionals, which may present not only with articular and physical symptomatology but also with increased anxiety symptomatology. In an attempt to systematize the different characteristics of JHS and comorbid symptoms, Bulbena et al. (4) have proposed a new phenotype, the 'neuroconnective' phenotype, based on core JHS and anxiety that is surrounded by five dimensions (behavioral, psychopathology, somatic symptoms, somatosensory symptoms and somatic illnesses), which includes several features of JHS in these areas. As we have shown, the core symptoms of JHS and anxiety are present from childhood, so further research should be carried out along these lines in children, with the aim of ensuring appropriate diagnosis and interventions. Although JHS often goes unnoticed and is considered, in most cases, a common benign

disorder, the features of the different areas involved may cause affected individuals significant difficulties. This impairment is especially relevant in children, who are in a fast-developing stage. JHS symptomatology may affect individuals through the psychological symptoms of anxiety, altering sleep patterns and self-esteem, reducing school performance and social and leisure activities, and possibly impacting on domestic life (30). Consequently, professionals should consider screening for anxiety symptomatology when treating high hypermobility.

There is evidence that the number of individuals identified with JHS is dependent on the criteria used to define JHS and the established cut-off (13, 17). Current criteria also need to prove their reliability and validity (44) and there is debate about their adequacy for pediatric populations (15, 45). A person-centered approach could allow us to overcome this drawback by identifying subgroups of children according to their similar hypermobility characteristics within groups and the differences between groups, in contrast to a variable-centered approach, which focuses on the description of the association between variables across individuals.

Hypermobility is common in childhood and tends to decline with age (13). The highest range of mobility was found in knee hyperflexion, the fifth finger, the shoulder and the patella. These frequencies are near to the results of Ohman (13) for a sample of a similar age to ours (5- o 8-year-old prepubertal children). Almost 40% ~~60%~~ of the children in the sample pertained to the high hypermobility class, indicating a high prevalence of hypermobility characteristics in 9 year-olds, which is in line with other reports (14). The musculoskeletal system is still developing in children and high hypermobility is expected until the stabilization of joint collagen with aging (46). As in other studies, there were no sex differences in hypermobility (12, 15).

This is the first study about the association of hypermobility and anxiety in children. The recruitment of a wide sample of the general population, the use of different reporters (clinicians observing the child and parents) and an analytical approach through a person-centered methodology are the strengths of the study. The results, however, should be interpreted considering some limitations. First, we did not use previously established criteria, but a selection of the hypermobility symptoms included in known criteria mostly reported in children to decide how they group empirically. Second, the internal consistency of parents' reports of physical injury fears scores was low, and this may compromise the interpretation of this result. Third, low SES families were the most reticent to participate in the study. However, we tested if there were any differences in anxiety and hypermobility by SES and found none. Fourth, due to the time spent with the children at schools being limited, we could not collect self-reports of anxiety. Fifth, there was no ongoing hypermobility assessment training to maintain quality assurance. However, the assessment was carried out in a relatively short period of time in a continuous manner and assessors were helped by pictures on the registration form of each child's evaluation.

The main finding of the study is to show that there is association between hypermobility and anxiety in children. This result has several clinical implications. Hypermobility and anxiety are both rooted in 'normality' and as such are characteristics that may be present to some extent in the population, so the first implication would be the diagnostic identification. The sample in this study was from the general population where absence of pathology was expected, which may explain the low effect sizes found. Even though there are statistically significant differences between the high/low LCA-Hypermobility groups on some of the anxiety measures, the mean in the high group is in the normal range for all of the anxiety scores. When anxiety is dichotomized,

between a 5.2% (obsessive compulsive) and a 18.5% (social phobia) of the children in the high hypermobility group showed elevated level of anxiety that would require further diagnostic examination. Literature has shown that children and adolescents with subclinical anxiety show greater individual and family psychopathology, increased risk for functional impairment (25, 47). Therefore, the results are informative and potentially relevant for clinicians. The results of the study suggest the need to screen for levels of anxiety among children high in JHS. If left undetected and untreated, anxiety may continue into adulthood and impair the daily functioning of the individual (48). The second implication would be to provide treatment if needed. One of the goals of the new neuroconnective phenotype proposal (JHS plus anxiety) mentioned above is for patients to receive comprehensive treatment. If anxiety levels are high, cognitive-behavior therapy (CBT) may be indicated alongside physical treatment. CBT is the election treatment for anxiety in children: some 60% of children recover following treatment (49). In adults with JHS plus anxiety, interventions involving the body, such as relaxation, postural techniques, aerobic exercise or strengthening, have also proved promising (8). Future research should test the efficacy of these approaches in children.

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Online materials

Table 1 online.

Comparison of high anxiety percentages between low/high LCA-Hypermobility groups

Figure 1.

Design of the Study

Table 1

Hypermobility criteria in the study

-
1. Passive dorsiflexion of the little fingers beyond 90° (one point if any hand)¹
 2. Passive apposition of the thumbs to the flexor aspects of the forearm (one point if any hand)¹
 3. Hyperextension of the elbows beyond 10° (one point if any elbow)¹
 4. External rotation of the shoulder up to more than 85° (one point if any shoulder)²
 5. Hypermobility of the patella (one point if any patella)²
 6. Hyperextension of the knee beyond 10° (one point if any knee)¹
 7. Hyperflexion of the knee allows to touch heel and buttock (one point if any knee)²
 8. Head rotation beyond 90° (one point if towards any side)
 9. Forward flexion of the trunk with knees fully extended so that the palms of the hands rest flat on the floor (one point)¹
-

¹Beighton criteria ²Hospital del Mar criteria

Table 2

Description of items of hypermobility and anxiety symptoms and comparison by sex

	Total sample (N = 336)	Girls (n = 162)	Boys (n = 174)	Sex comparison <i>p</i>
<i>Hypermobility - % of presence</i>				
Fifth finger	75.6	73.5	77.5	.395
Thumb forearm	59.1	57.9	60.2	.665
Elbow	59.6	58.2	60.8	.621
Shoulder	74.0	76.9	71.3	.244
Patella	73.4	74.3	72.6	.722
Knee hyperextension	35.9	37.4	34.5	.592
Knee hyperflexion	81.8	83.6	80.2	.428
Head	68.4	70.5	66.4	.417
Trunk	39.3	51.5	27.9	<.001
Total Score – mean (SD)	5.67 (1.9)	5.84 (1.9)	5.51 (1.9)	.113
<i>Spence Children's Anxiety Scale- Parent version – mean¹ (SD)</i>				
<i>[Percentil 5 – Percentil 95 scores]</i>				
Separation anxiety	2.78 (2.4) [0.0 - 7.5]	2.89 (2.4) [0.0 - 7.2]	2.68 (2.5) [0.0 - 8.0]	.409
Social phobia	3.91 (2.6) [0.0 - 9.0]	4.17 (2.8) [0.0 - 9.2]	3.66 (2.5) [0.0 - 9.1]	.071
Obsessive compulsive	0.47 (1.2) [0.0 - 2.0]	0.53 (1.1) [0.0 - 2.7]	0.42 (1.3) [0.0 - 2.0]	.373
Panic/agoraphobia	0.45 (1.2) [0.0 - 3.0]	0.44 (0.9) [0.0 - 3.0]	0.46 (1.4) [0.0 - 2.0]	.885
Physical injury fears	2.12 (2.0) [0.0 - 6.0]	2.36 (2.2) [0.0 - 7.0]	1.89 (1.8) [0.0 - 5.7]	.035
Generalized anxiety	2.53 (2.17) [0.0 - 6.0]	2.51 (2.2) [0.0 - 6.2]	2.55 (2.0) [0.0 - 6.0]	.845
Total Score	12.24 (8.5) [2.0 - 29.0]	12.88 (8.4) [3.0 - 31.2]	11.65 (8.6) [2.0 - 26.0]	.183

In bold *p* < .05. ¹ Mean raw scores

Table 3.

Latent Class Analysis solutions from 9 hypermobility items

# Latent classes	AIC	BIC	Lo-Mendell Rubin (<i>p</i>)	Class: N	Class: probability*	Entropy
1c	3621.9	3656.0	-	1: 329	-	-
2c	3520.9	3593.0	.002	1: 135 2: 201	1: .839 2: .937	.644
3c	3497.7	3607.8	.510	1: 68 2: 133 3: 135	1: .721 2: .832 3: .870	.625
4c	3468.4	3616.4	.065	1: 140 2: 60 3: 70 4: 66	1: .909 2: .932 3: .947 4: .907	.855
5c	3471.6	3657.6	.852	1: 142 2: 11 3: 58 4: 59 5: 60	1: .922 2: .828 3: .925 4: .858 5: .892	.845

* Posterior probability of class membership

In bold selected solution of LCA

Table 4.

Comparison of hypermobility and anxiety symptoms between low/high LCA-hypermobility groups

	Low (n = 201)	High (n = 135)	p	RR
<i>Hypermobility - % of presence</i>				
Fifth finger	63.3	93.7	<.001	1.48
Thumb forearm	31.6	100.0	<.001	3.16
Elbow	46.8	78.5	<.001	1.68
Shoulder	61.3	92.9	<.001	1.52
Patella	57.0	97.8	<.001	1.72
Knee hyperextension	18.9	61.2	<.001	3.24
Knee hyperflexion	81.3	82.6	.761	1.02
Head	58.3	83.4	<.001	1.43
Trunk	35.1	45.7	.055	1.30
Total Score – mean (SD)	4.54 (1.4)	7.35 (1.0)	<.001	2.29
<i>Spence Children's Anxiety Scale - Parent version – mean¹ (SD)</i>				
<i>[Percentil 5 – Percentil 95 scores]</i>				<i> d </i>
Separation anxiety	2.48 (2.3) [0.0 - 7.0]	3.22 (2.6) [0.0 - 8.2]	.007	0.31
Social phobia	3.60 (2.6) [0.0 - 9.0]	4.35 (2.6) [1.0 - 10.0]	.010	0.29
Obsessive compulsive	0.46 (1.0) [0.0 - 2.0]	0.49 (1.4) [0.0 - 2.8]	.811	0.02
Panic/agoraphobia	0.36 (0.8) [0.0 - 2.0]	0.58 (1.6) [0.0 - 3.0]	.135	0.17
Physical injury fears	1.89 (1.9) [0.0 - 6.0]	2.45 (2.2) [0.0 - 7.0]	.013	0.28
Generalized anxiety	2.35 (2.1) [0.0 - 6.0]	2.80 (2.1) [0.0 - 6.0]	.053	0.22
Total Score	11.13 (7.9) [2.0 - 29.0]	13.90 (9.1) [3.0 - 28.9]	.003	0.33

In bold $p < .05$. RR: Risk ratio; d: Cohen's d . ¹ Mean raw scores