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RESEARCH ARTICLE

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Hypermobility, developmental coordination disorder and physical activity in an Irish paediatric population

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Abstract

Objective: The primary aim of the study was to explore the prevalence of generalized joint hypermobility (GJH) and generalized hypermobility spectrum disorder (gHSD) using the new classification system in a community paediatric physiotherapy service in Ireland. The second aim was to explore the relationship between GJH, gHSD and physical activity level, while considering the association of probable developmental coordination disorder (pDCD).

Methods: A case-controlled cross-sectional study of children aged 6–12 years, recruited from the community paediatric physiotherapy department (n = 32) and a local school (n = 41), was carried out. A Beighton score of ≥6/9 distinguished GJH. The new framework for hypermobility spectrum disorder (HSD) was used. Self-reported physical activity level was measured using the Physical Activity Questionnaire—Older Children. A parent-reported validated questionnaire screened for pDCD.

Results: The prevalence of GJH was 21.9% of children attending physiotherapy. One child in the physiotherapy group was identified as having gHSD, with a prevalence of 3.1%. There was no significant difference in physical activity level between children with and without GJH attending physiotherapy (independent samples t-test, p = 0.28). Probable developmental coordination disorder (pDCD) was observed in 71.9% of children attending physiotherapy. There was no significant difference in the number of children with pDCD in those with and without GJH (Fisher's exact test, p = 0.370).

Conclusions: This study was the first to explore the prevalence of GJH and gHSD in the paediatric physiotherapy population in Ireland. The presence of GJH did not affect self-reported physical activity level or motor coordination in children attending physiotherapy.

KEYWORDS

musculoskeletal, paediatrics, physiotherapy, generalized joint hypermobility, generalized hypermobility spectrum disorder, physical activity, developmental coordination disorder, motor coordination

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1 | INTRODUCTION

Joint hypermobility (JH) is defined as an excessive range of motion of a joint, taking into consideration the patient's gender, age and ethnic background (Grahame, 2009). Generalized joint hypermobility (GJH) is often asymptomatic, and can even be an asset in terms of sports performance—for example, in ballet or gymnastics (Grahame, 2009). Some children with GJH present with symptoms, with pain in multiple joints, fatigue, skin hyperextensibility or fragility, and unstable joints being the most common (Pacey, Adams, Tofts, Munns, & Nicholson, 2015; Pacey, Tofts, Adams, Munns, & Nicholson, 2015). Before 2017, children with these symptoms were classified as having joint hypermobility syndrome (JHS) or, in some cases, Ehlers–Danlos syndrome–hypermobility type (EDS-HT). Many experts suggested that JHS and EDS-HT were indistinguishable, and this led to confusion in terminology. In 2017, as a result of an international expert consensus group, a new framework and nomenclature for the hypermobility spectrum in children and adults was proposed. In this new framework, when JH occurs together with symptoms (usually musculoskeletal), and when hypermobile Ehlers–Danlos syndrome (hED) or other Ehlers–Danlos syndromes have been excluded, it is referred to as hypermobility spectrum disorder (HSD) (Castori et al., 2017). This new classification for HSD, which is now internationally acknowledged, describes four different phenotypes of HSD: localized HSD, historical HSD, peripheral HSD and generalized HSD (gHSD).

The cohorts of interest in the present study were those with GJH and gHSD. In the paediatric population, the upper cut-off point for the Beighton score is 6/9 when identifying GJH (Juul-Kristensen et al., 2009). The Beighton score is calculated through bilateral measurement of passive dorsiflexion of the fifth metacarpal joint; passive apposition of the thumb to the forearm; hyperextension of the elbow; hyperextension of the knee and forward flexion of the trunk. The prevalence of GJH (Beighton score $\geq$6/9) in Danish and Dutch children aged 6–12 years has been reported to range from 9.4% to 35.0% (Juul-Kristensen et al., 2009; Remvig, Kummel, Halkjaer Kristensen, Boas, & Juul-Kristensen, 2011; Smits-Engelsman, Klerks, & Kirby, 2011). To date, there have been no data for Irish schoolchildren. Before 2017, the Brighton criteria were used to diagnose JHS (Grahame, Bird, & Child, 2000). JHS and gHSD are comparable, as they both describe individuals with symptomatic JH. Two Danish studies assessed schoolchildren (aged 8 years and 10 years, respectively) for JHS using the Brighton criteria, and reported a prevalence of 17.6% and 9%, respectively (Juul-Kristensen et al., 2009; Remvig et al., 2011). Castori et al. (2017) described gHSD as the presence of GJH, measured by a Beighton score of $\geq$6/9, plus one or more secondary musculoskeletal manifestations. No prevalence data have been collected using the new framework for the diagnosis of gHSD.

Physical activity is defined as "any bodily movement that expends energy" (World Health Organization, 2017). Physical activity assists children and young people to develop their musculoskeletal system, cardiovascular system and neuromuscular awareness, and maintain a healthy body weight (World Health Organization, 2017). It has been suggested that limitations imposed by GJH can have an impact on physical activity levels due to symptoms associated with the disorder, such as pain and fatigue (Engelbert, van Bergen, Henneken, Helders, & Takken, 2006; Tobias, Deere, Palmer, Clark, & Clinch, 2013). However, the current literature suggests that there are no significant differences in physical activity level in children with and without GJH (Juul-Kristensen et al., 2009; Leone et al., 2009; Qvindesland & Jonsson, 1999; Remvig et al., 2011). Further investigation into JHS and physical activity levels has resulted in conflicting findings. Self-reported physical activity level has been shown to be reduced in children with JHS in a healthcare setting, whereas there was no difference in self-reported physical activity level in Dutch children in a school setting (Engelbert et al., 2006; Juul-Kristensen et al., 2009; Remvig et al., 2011; Schubert-Hjalmarsson, Ohman, Kyllerman, & Beckung, 2012). However, it is well accepted that self-reported measures of physical activity have been shown to have low to moderate correlation with direct measures of physical activity (e.g., accelerometry) in the paediatric population, so these results should be interpreted with caution (Adamo, Prince, Tricco, Connor-Gorber, & Tremblay, 2009).

Developmental coordination disorder (DCD), also known as dyspraxia, is a common disorder affecting gross and/or fine motor coordination in children and adults (Blank, Smits-Engelsman, Polatajko, & Wilson, 2011). DCD and JHS have many overlapping features, such as impaired proprioception, coordination difficulties, low muscle tone and joint pains (Adib, Davies, Grahame, Woo, & Murray, 2005; Kirby & Davies, 2007; Pacey, Adams, Tofts, Munns, & Nicholson, 2014; Rombaut et al., 2012; Scheper et al., 2016; Smith et al., 2013). Neurodevelopmental problems, JH and hypermobility spectrum disorders are a growing area of interest, as they might explain a subgroup of people who struggle with body awareness (Bulbena-Cabré et al., 2017). Kirby and Davies (2007) reported a significant increase in the prevalence of JHS in children with DCD (37%) vs. typically developing children (7.4%) ($p < 0.05$) (Kirby & Davies, 2007). These authors used the five-point questionnaire to measure JH, which is more commonly used in the adult population and has not been validated for the paediatric population (Juul-Kristensen, Schmedling, Rombaut, Lund, & Engelbert, 2017).

The diagnostic criteria for DCD are defined in the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV) (American Psychiatric Association, 2000). The prevalence of DCD is 5–6% in 6–11-year-olds (American Psychiatric Association, 2013). Motor-based questionnaires and measures that fall outside the DSM-IV do not verify a diagnosis of DCD but identify subjects with probable DCD (pDCD) (Blank et al., 2011). The Developmental Coordination Disorder Questionnaire 2007 (DCDQ'07) is a measure designed to screen for pDCD in children aged 5–15 years (Wilson et al., 2009). There is evidence that children with DCD and pDCD have reduced physical activity levels (Cairney et al., 2005; Cantell & Crawford, 2008; Oudenampsen et al., 2013; Rivilis et al., 2011; Silman, Cairney, Hay, Klentrou, & Faught, 2011; Visser, Geuze, & Kalverboer, 1998).

The primary aim of the present research was to explore the prevalence of GJH and gHSD using the new classification system in a community paediatric physiotherapy service in Ireland. The second aim

was to explore the relationship between GJH, gHSD and physical activity level, while also considering the association of pDCD.

2 | METHODS

2.1 | Study design, sampling and setting

A cross-sectional, case-controlled, observational study design was used to compare children referred to a community paediatric physiotherapy services with a comparison group of children recruited from a local primary school. The physiotherapy group were children aged 6–12 years who had been referred to an Irish community paediatric physiotherapy department. Children who received an initial physiotherapy appointment letter from mid-April to June 2017 were invited to take part in the study. Children were excluded if they had a known hereditable connective tissues disorder or neurological impairment. The comparison group or school group were children aged 6–12 years, recruited from a nearby primary school using convenience sampling. Ethical approval was sought from the Clinical Research Ethics Committee of the Cork Teaching Hospitals (CREC), reference number ECM 4(ii).

2.2 | Questionnaires

Children who consented to participate in the study received a pack by post, containing self-reported questionnaires. Children completed the Physical Activity Questionnaire for older Children (PAQ-C). The PAQ-C is a self-administered 7-day recall instrument measuring activity level during physical education, school break/lunch, after school and at the weekend.

Parents or guardians also completed the DCDQ'07 screening tool. An additional research-specific questionnaire was completed by the parent/guardian and child, providing information on chronic joint pain (lasting >3 months in at least one joint) and joint dislocation/subluxation, in order to detect secondary musculoskeletal manifestations and enable identification of gHSD.

2.3 | Physical assessment

The physical assessment involved measurements of the child's height, weight and Beighton score to identify GJH using standardized procedures. Smits-Engelsman et al. (2011) and have documented standardized protocols for the Beighton score, and these were used in the present study. Passive dorsiflexion of the fifth metacarpal joint, hyperextension of the elbow and hyperextension of the knee were measured using a plastic two-legged 360-degree goniometer (type HIRes (Baseline Evaluation Instruments, USA)). The physical assessment was part of the standard initial community paediatric physiotherapist assessment. Two researchers visited the school during April 2017 to complete the physical assessments in the school group. The researcher with a background in paediatric physiotherapy completed all the physical assessments. Both of the researchers were blinded to the results of the self-reported questionnaires (Figure 1).

2.4 | Data analysis and statistics

Data analysis was performed using SPSS version 22.0 (IBM, Armonk, New York, USA). Descriptive statistics, including mean, median, standard deviation (SD), interquartile ranges (IRQs), frequencies and percentages were used to summarize participant characteristics. Body mass index z-scores (BMIz) were calculated using height and weight, and compared with the World Health Organization interpretation of cut-offs for BMI z-scores in children aged 5–19 years (World Health Organization, 2007). All variables were checked for normality. Data were examined for any differences between groups, using the independent-samples t-test for normally distributed continuous data, the Mann–Whitney U-test for non-normally distributed data and the chi-square test for categorical data. Fisher's exact test was selected to compare the difference in prevalence and gender between two groups when the assumptions for the chi-square test were not met—that is, the lowest expected frequency in any cell was <5 (Yates, Moore, & McCabe, 1999). The Spearman's rank correlation coefficient was selected to measure the strength of correlation between two groups when either one or both variables were non-normally distributed. A correlation coefficient of $\geq$0.10 was considered to represent a weak association, $\geq$ 0.30 represented a moderate association and $\geq$0.50 represented a strong association (Cohen, 1992). Significance was determined at the 0.05 level for all measures.

3 | RESULTS

A total of 48 children attending physiotherapy and 57 local schoolchildren were invited to take part in the study. Only full data sets were included in the statistical analysis. Study participants comprised 32 children attending physiotherapy and 41 local schoolchildren (Figure 2). Demographic data are displayed in Table 1.

Sixty-three children were of normal weight (86.3%), one child was underweight (1.4%), five were overweight (6.8%) and four were obese (5.5%), applying the World Health Organization interpretation of cut-offs for BMI z-scores in children aged 5–19 years (World Health Organization, 2007). Of the physiotherapy group, 32.3% (n = 10/32) reported lower limb pain, 12.5% (n = 4/32) upper limb pain, 16.1% (n = 5/32) neck/back pain and 6.5% (n = 2/32) headaches. Of the school group, 13.5% (n = 5/41) reported lower limb pain, 7.3% (n = 3/41) upper limb pain and 2.4% (n = 1/41) back pain.

3.1 | Prevalence of GJH

A Beighton score of $\geq$6/9 was observed in 21.9% (n = 7/32) of the physiotherapy group and 17.1% (n = 7/41) of the school group (Table 2). There was no significant difference in the prevalence of GJH between the two groups (chi-square test, p = 0.605). There was a higher prevalence of GJH in girls (28.0%) vs. boys (14.6%) but this

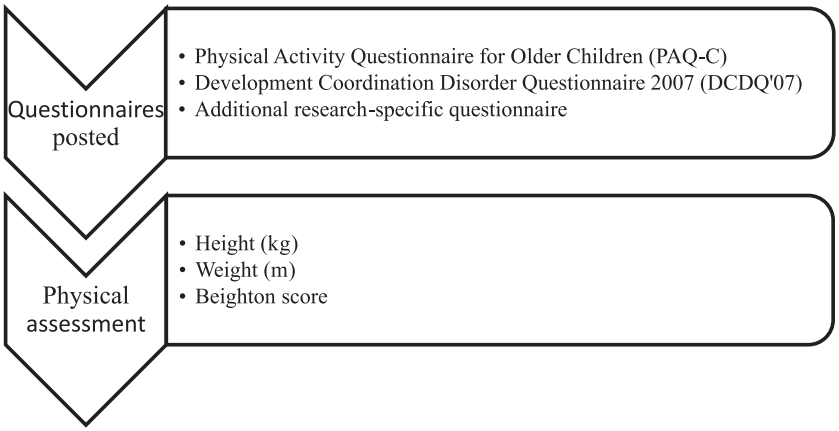

FIGURE 1 Flowchart of study procedure

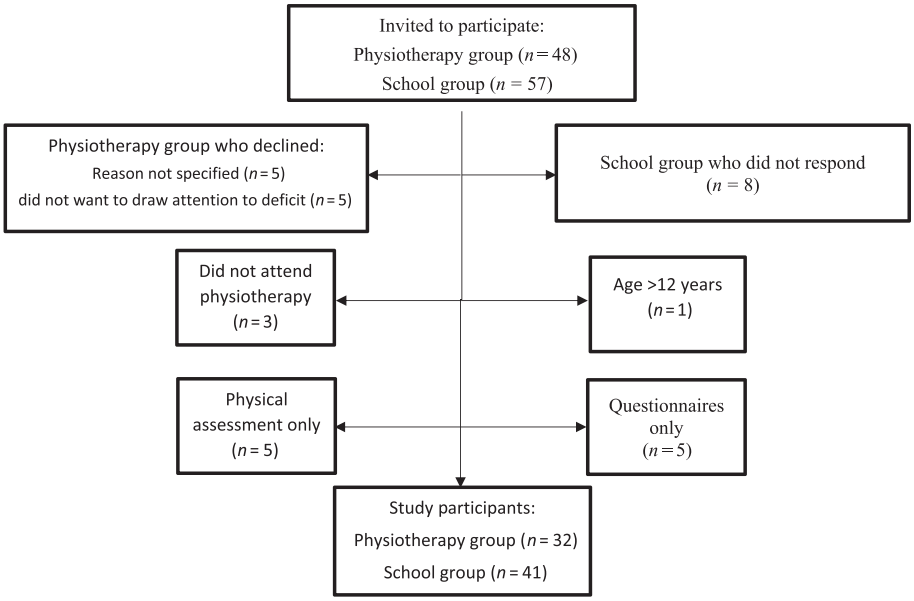

FIGURE 2 Flow diagram of participant recruitment

TABLE 1 Demographics of school group and physiotherapy group

		Physiotherapy group (*n* = 32)	School group (*n* = 41)	*p*-Value
Gender	Girls, *n* (%)[a] Boys, *n* (%)	6 (19) 26 (81)	19 (46) 22 (54)	0.014*
Ethnicity	Caucasian, *n* (%) Non-Caucasian, *n* (%)	32 (100) 0 (0)	40 (98) 1 (2)	– –
Age in years, median (IQR)[b]		8.6 (2.5)	10.7 (3.6)	<0.005*
Height in cm, mean (SD)[c]		135.8 (12.6)	144.5 (13.4)	0.238
Weight in kg, mean (SD)[c]		33.6 (9.6)	40.3 (12.3)	0.126
BMI in kg/m^2, mean (SD)[c]		17.9 (3.1)	18.6 (3.0)	0.508

[a]Chi-square test.

BMI: body mass index; IQR: interquartile range; SD: standard deviation.

*$p < 0.05$.

[b]Mann–Whitney U-test.

[c]Independent-samples t-test.

TABLE 2 Beighton score for both participant groups

Beighton score	Physiotherapy group (*n* = 32), *n* (%)	School group (*n* = 41), *n* (%)
9	0 (0)	2 (4.9)
8	2 (6.3)	1 (2.4)
7	0 (0)	1 (2.4)
6	5 (15.6)	3 (7.3)
	Cut-off for GJH using Beighton score ≥6/9	
5	1 (3.1)	1 (2.4)
4	9 (28.1)	9 (22.0)
3	3 (9.4)	3 (7.3)
2	7 (21.9)	8 (19.5)
1	0 (0)	2 (4.9)
0	5 (15.6)	11 (26.8)

GJH, generalized joint hypermobility

did not reach statistical significance (Fisher's exact test, $p = 0.214$). The most frequently recorded hypermobile joints were the left elbow ($n = 37$) and the right knee ($n = 37$), followed by the left knee ($n = 36$) and right elbow ($n = 35$) (Figure 3).

3.2 | Prevalence of gHSD

One child in the physiotherapy group met the criteria for gHSD, with a prevalence of 3.1%. This participant was a 7-year-old Caucasian boy presenting to physiotherapy for the first time with neck pain, fatigue, reduced balance and low muscle tone (Table 3). He had a Beighton score of 6/9. None of the children in the school group met the criteria for gHSD.

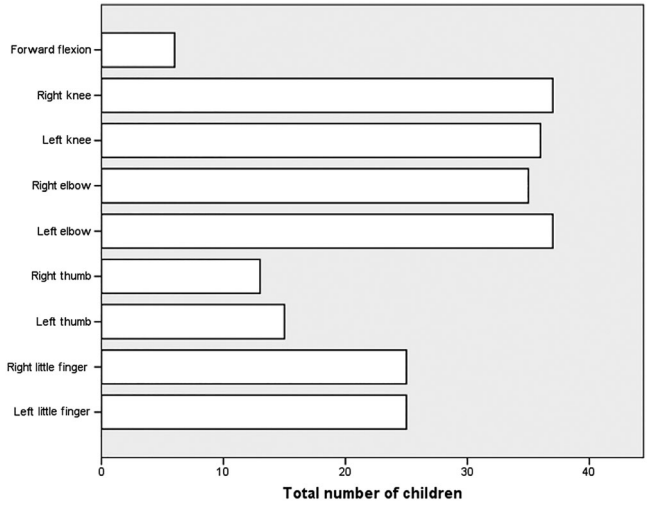

FIGURE 3 Number of hypermobile joints, as measured using the Beighton score in all participants ($n = 73$)

3.3 | Relationship between GJH, gHSD and physical activity level

As only one child met the criteria for gHSD in the physiotherapy group and entire study sample, the sample size was insufficient to compare the physical activity level of children with and without gHSD. However, the data did allow for comparison of scores between those with and without GJH in the physiotherapy and school groups (Table 3).

There was no significant difference in the physical activity level between children with (mean [SD] 2.8 [0.7]) and without (3.1 [0.7]) GJH in the physiotherapy group (independent-samples t-test, $p = 0.50$). There was also no significant difference in the physical activity level between children with (mean [SD] 3.7 [0.5]) and without (3.7 [0.5]) GJH in the school group (independent-samples t-test, $p = 0.93$). Of interest, the physical activity level was significantly lower in the physiotherapy group (mean [SD] 3.0 [0.7]) compared with the school group (3.7 [0.5]) (independent-samples t-test, $p < 0.005$) (Figure 4).

A PAQ-C cut-off of 2.87 discriminates between those meeting physical activity guidelines and those who do not (Voss, Dean, Gardner, Duncombe, & Harris, 2017). This cut-off is based on meeting physical activity guidelines of ≥60 min per day. Of the physiotherapy group, 43.8% ($n = 14/32$) and 73.2% ($n = 30/41$) of children in the school group met the national guidelines for physical activity in Ireland. The child with gHSD scored 2.58 on the PAQ-C and did not meet the national guidelines for physical activity.

3.4 | Prevalence of pDCD

pDCD was observed in 71.9% ($n = 23/32$) of the physiotherapy group and 22.0% ($n = 9/41$) of the school group. There was a significantly higher prevalence of pDCD in the physiotherapy group (chi-square test, $p < 0.005$).

There was no significant difference in the number of children with pDCD in those with ($n = 4/7$, 57.1%) and without ($n = 19/25$, 76%) GJH in the physiotherapy group (Fisher's exact test, $p = 0.370$). There was also no significant difference in the number of children with pDCD in those with ($n = 2/7$, 28.6%) and without ($n = 7/34$, 20.6%) GJH in the school group (Fisher's exact test, $p = 0.637$).

There was a moderate positive correlation between DCDQ'07 score and PAQ-C score in the physiotherapy group (Spearman's $r_s = 0.32$, $p = 0.13$) and a significant moderate positive correlation in the school group (Spearman's $r_s = 0.47$, $p = 0.01$). A higher DCDQ'07

TABLE 3 Physiotherapy group referring complaint

Referring complaint	Frequency	
	GJH (*n* = 7)	Non-GJH (*n* = 25)
Acute soft tissue injury	3	10
Lower limb alignment	1	6
Flat feet	1	3
Low muscle tone	1	2
Chronic joint pain	1	4

GJH, generalized joint hypermobility

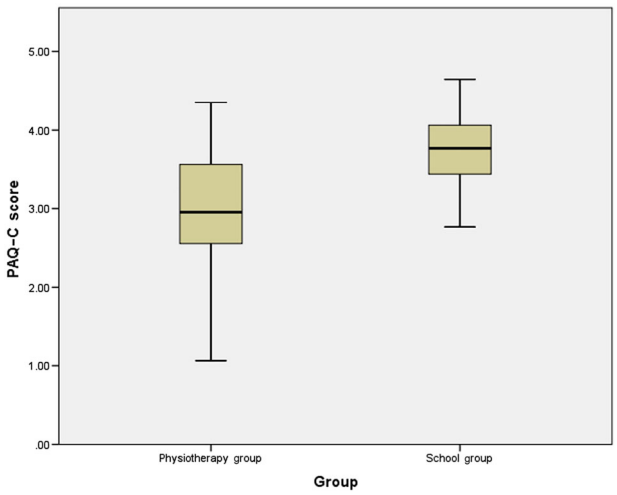

FIGURE 4 Physical activity level in the Physiotherapy group compared with the school group. A Physical Activity Questionnaire for older Children (PAQ-C) activity summary score of 1 indicates low physical activity, whereas a score of 5 indicates high physical activity

score was indicative of enhanced motor coordination, and a higher PAQ-C score was indicative of greater physical activity level (Table 4).

4 | DISCUSSION

The present study demonstrated a prevalence of GJH of 21.9% in children attending a paediatric physiotherapy department and 17.1% in local schoolchildren in Ireland. These figures are comparable with the results from a previous study of Dutch schoolchildren of the same age (23.0%) but are slightly higher than in the data obtained from schoolchildren in Denmark (9.4–11.1%) using a Beighton score ≥6/9 (Juul-Kristensen et al., 2009; Remvig et al., 2011; Smits-Engelsman et al., 2011). One of these children presented with chronic joint pain lasting more than 3 months, reinforcing the idea of GJH as a normal variant (Remvig, Jensen, & Ward, 2007).

The overall prevalence of gHSD in 6–12-year-old children attending physiotherapy was 3.1% using the new framework for HSD. There are no known data for the prevalence of gHSD in children attending physiotherapy, and none of the children in the school group met the criteria for gHSD. This figure is much lower than previously reported for JHS/EDS-HT using the Brighton criteria, where prevalences of 8.9% (Juul-Kristensen et al., 2009) and 17.6% (Remvig et al., 2011) were reported in schoolchildren aged 8–10 years from mainland Europe. Results from this study agree with those in previous literature which proposed that JHS/EDS-HT may have been over-diagnosed in the paediatric population (Junge, Jespersen, Wedderkopp, & Juul-Kristensen, 2013; Remvig et al., 2011; Smits-Engelsman et al., 2011). Castori et al. (2017) have raised the threshold for identification of gHSD. Not all children diagnosed with JHS/EDS-HT in the past will now meet the description of gHSD. These children and their parents will no longer have their symptoms validated by the health profession. This may hinder access to clinical treatment, tertiary services, resource hours or assistance in school. These results should be viewed with caution owing to potential recruitment bias; for example, one child previously diagnosed with JHS/EDS-HT declined to take part in the study. A further five parents declined to take part as they did not want to discuss physical ability and draw attention to the child's deficits. These were potentially children with JH and pain who were opting out.

The work of Castori et al. (2017) has resulted in clearer diagnostic criteria for hEDS. However, there are no clear criteria for gHSD, which is a diagnosis of exclusion, whereby individuals with symptomatic GJH that do not satisfy the criteria for a hereditary connective tissue disorder are diagnosed with gHSD. It is therefore up to research teams to develop their own strict inclusion and exclusion criteria. For example, children have been excluded with any other pathological condition with hypermobility as a known feature, a current or previous orthopaedic condition and a diagnosis of neurodevelopmental disorders (Adib et al., 2005; Ferrari et al., 2005; Junge et al., 2013; Pacey, Tofts, Adams, Munns, & Nicholson, 2015). The fact that this varies from study to study limits the ability to compare data directly. However, the new framework for HSD is intended as a guide for health professionals, based on current available literature. It allows the clinician a degree of flexibility rather than a rigid policy (Castori et al., 2017).

The children in the present study were considered symptomatic if they reported joint pain lasting >3 months or recurrent joint dislocations or subluxations. Castori et al. (2017) suggested other secondary musculoskeletal features indicative of HSD, including disturbed proprioception, muscle weakness, and pelvic and bladder dysfunction. As these factors are much more difficult to quantify, they were not investigated in the present study. In addition, the author of this study did not have access to medical records, and relied on the parent to recall medical history; therefore, children with proprioception or bladder dysfunction were not identified in the present study. The diagnosis

TABLE 4 Results from self-report questionnaires for physical activity and probable DCD

	Physiotherapy group			School group		
	GJH (*n* = 7)	Non-GJH (*n* = 25)	Total (*n* = 32)	GJH (*n* = 7)	Non-GJH (*n* = 34)	Total (*n* = 41)
PAQ-C, mean (SD)	2.8 (0.7)	3.1 (0.7)	3.0 (0.7)	3.7 (0.5)	3.7 (0.5)	3.7 (0.5)
DCDQ'07, median (IQR)	35.5 (22.5)	43.5 (20.8)	42.0 (22.3)	61.0 (24.0)	66 (15.0)	65.5 (17.5)

The PAQ-C summary score is marked out of 5; a score of 1 indicates low physical activity, and a score of 5 indicates high physical activity. The DCDQ'07 is marked out of 100; higher scores indicate better motor coordination. DCD: developmental coordination disorder; DCDQ'07: Developmental Coordination Disorder Questionnaire 2007; GJH: generalised joint hypermobility; IQR: interquartile range; PAQ-C: Physical Activity Questionnaire for Older Children; SD: standard deviation.

and classification of HSD and hEDS are an ongoing process and will need to be further clarified for clinical research teams.

There was a higher prevalence of GJH in the physiotherapy group (n = 7/32, 21.9%), and although only one child reported joint pain lasting >3 months, three children presented with acute soft tissue injuries. Tobias et al. (2013) identified GJH (Beighton score ≥6/9) at a mean age of 13.8 years as a statistically significant risk factor for musculoskeletal pain at mean age of 17.8 years. El-Metwally, Salminen, Auvinen, Kautiainen, and Mikkelsson (2005) reported that GJH (Beighton score ≥6/9) was predictive of lower limb pain recurrence at the 4-year follow-up, compared with those with normal joint laxity, in Finish schoolchildren. It would be of interest to follow the cohort from this study for the longer term and investigate whether or not they develop persistent joint pain in the future, as suggested by Tobias et al. (2013) and El-Metwally et al. (2005).

There was a significantly higher prevalence of pDCD in the physiotherapy group (78.6%) than in the school group (26.5%). These figures were much higher than the estimated prevalence of DCD in children, at 5–6% in 5–11-year-olds, using the DSM-IV criteria, which include standardized objective measures (Blank et al., 2011). This was somewhat expected, as the DCDQ'07 is a screening tool which is inherently more likely to over-identify children than to miss a child who has motor coordination problem. The DCDQ'07 may have identified children who did not have the condition, but further standardized motor testing using the DSM-IV criteria would have revealed whether DCD was indeed present; however, this was beyond the scope of the current research.

Of interest, four children (57.1%) in the physiotherapy group with a Beighton score of ≥6/9 also presented with pDCD. Using the old Beighton score cut-off of ≥4/9, the number of children with pDCD went up to 12 (70.6%). If gHSD and pDCD were truly overlapping, as suggested in the literature, then one might expect the proportion of pDCD to be higher in the children with more hypermobile joints—that is, scoring ≥6/9(Kirby & Davies, 2007). In fact, the proportion of children with pDCD reduced using the higher cut-off of ≥6/9.The data from the present study suggested that hypermobility and pDCD should be considered as separate entities. Having more hypermobile joints was not associated with having impaired motor control. As a result of the limited sample size and the use of a subjective measure of motor control, it was impossible to draw firm conclusions from the current data set, but this highlights an area for future research.

The one child diagnosed with gHSD did not meet recommended physical activity guidelines. There was no significant difference in physical activity level in children with or without GJH in either the school group or the physiotherapy group. These data are in agreement with those from a comparable cohort of Danish schoolchildren, aged 8-10 years, where there was also no significant difference in physical activity level in children with and without GJH using a Beighton score cut-off of ≥6/9 (Juul-Kristensen et al., 2009; Remvig et al., 2011).

There was a moderate positive correlation between the DCDQ'07 score and PAQ-C score in both the physiotherapy group and the school group. In other words, children with pDCD reported lower physical activity levels. This is in agreement with the current literature base in children aged 7–14 years (Cairney et al., 2005; Cantell & Crawford, 2008; Oudenampsen et al., 2013; Silman et al., 2011; Visser et al., 1998). However, these five studies used different measures of DCD or pDCD and physical activity, so a direct comparison with these data could not be undertaken. Inaccuracies in physical activity level questionnaires are particularly reported in paediatric populations, and children have a tendency to over-report their activity levels (Adamo et al., 2009). The PAQ-C scores collected from this sample should be viewed with caution as they are most likely to be an overestimation of the actual physical activity undertaken. The results of the current study add to the evidence suggesting a developing trend for lower physical activity level in children with reduced motor competence, but further investigation into this relationship is required.

4.1 | Limitations

The current study utilized a convenience sample of children. The sample size was too small to recruit a sufficient number of children with gHSD for meaningful statistical analysis on the original study aims to explore the relationship between gHSD and physical activity level. Use of the PAQ-C and DCDQ'07, which rely on recall and self-reporting, limits the ability to draw firm conclusions. Use of the DSM-IV criteria to diagnose DCD and an objective measure of physical activity would increase the strength of any future research. It would be of interest to follow this cohort over a longer period, to investigate whether or not these children develop chronic joint pain and how their physical activity level alters with increasing age. With clarification of the Beighton score cut-off for children, and the new classification for gHSD, further exploration of the overlapping features of DCD is warranted.

5 | CONCLUSION

Using the new framework for diagnosis in hypermobility, the prevalence of gHSD in children attending physiotherapy was 3.1% (n = 1/32). The prevalence of GJH was 21.9% (n = 7/32) in children attending physiotherapy. There was no significant difference in physical activity level or the proportion of children presenting with pDCD in those with and without GJH in 6–12-year-olds attending physiotherapy. Inclusion of the DSM-IV criteria to diagnose DCD and an objective measure of physical activity would increase the strength of any future research. The new HSD framework and higher cut-off for GJH may begin to tease out the overlapping features of gHSD and DCD in the clinical setting. This warrants further exploration in much larger samples.

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CONFLICTS OF INTEREST

The authors declare that there are no known conflicts of interest associated with any of the authors.

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