

## Association between Joint Hypermobility Syndrome and Developmental Coordination Disorder – A Review

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### Abstract

**Introduction:** The term joint hypermobility syndrome (JHS) was adopted after clinicians became aware of the myriad of symptoms associated with this multisystemic condition. JHS is an inherited disorder of connective tissues affecting the musculoskeletal and visceral systems which may contribute to a reduction in health related physical fitness. Pain associated with JHS may be influenced by hypermobility and biomechanical dysfunction. Biomechanical dysfunction observed in patients with JHS may be as a result of impaired motor control and in particular developmental coordination disorder (DCD). DCD (described in the literature utilising the terms clumsy child syndrome; perceptual motor dysfunction; dyspraxia) is a neurodevelopmental condition characterised by coordination difficulties affecting function. The objective of this review is to examine the association between hypermobility, JHS, motor control impairment and DCD.

**Methods and data sources:** EMBASE, MEDLINE, CINAHL, ASSIA, PsychARTICLES, SPORTDiscus and PsycINFO from 1989 - 2009. Research articles written in English and peer reviewed were included.

**Results:** Five research papers were identified. The studies employed a variety of methodologies and assessment tools for reporting joint hypermobility, JHS, motor delay, motor impairments and DCD. All five studies reported on children between the ages of six months and 12 years. Three out of four studies reported on association between impaired motor development, motor delay and joint hypermobility. There was no consensus as to whether motor delay, impaired motor development and joint hypermobility continued as the child matured. One study ascertained that children with JHS reported similar functional difficulties as children with DCD.

**Conclusion:** There was a paucity of literature relating to an association between joint hypermobility, JHS, impaired motor control, motor delay and DCD in children, there was no literature pertaining to adults. This association requires further exploration if professionals are to understand, nurture and manage those reporting these associated conditions.

**Keywords:** Motor control impairments; Coordination; Hypermobility; Musculoskeletal pain; Motor delay; Athletes

**Abbreviations:** BJHS: Benign Joint Hypermobility Syndrome; DCD: Developmental Coordination Disorder; DSM: Diagnostic Statistical Manual of Mental Disorders; EDS: Ehlers-Danlos Syndrome; JHS: Joint Hypermobility Syndrome; MFS: Marfan Syndrome; OI: Osteogenesis Imperfecta; VMI: Visual Motor Integration

### Introduction

Hypermobility is frequently recognised in athletes and performing artistes [1-4], is associated with increased rates of injury and prolonged rehabilitation [2,4]. It may be observed globally, unilaterally or in one or a few joints [5-7]; the joints commonly affected are sport or activity dependent [3,8]. Hypermobility is demonstrated where there is excessive range of movement. Hypermobility recorded in population studies has been found to be more prevalent in females than males [5,9] where it is possible that hormonal influences may contribute to increased laxity in women [10]. There is a higher prevalence of hypermobility amongst Asians and Africans than Caucasians [11,12]. It is reported to decline with age but anecdotal evidence suggests many with hypermobility retain increases in range of movement enabling continued agility and activity in later life [11,13].

Hypermobility and musculoskeletal symptoms in the absence of rheumatic disease were first described by Kirk and colleagues in 1967 [14]. The musculoskeletal symptoms described were overuse injuries, joint pain and recurrent dislocations. The term joint hypermobility syndrome (JHS) was adopted after clinicians became aware of the myriad of symptoms associated with this multisystemic condition [7]. JHS is one of the inherited disorders of connective tissues and shows symptom overlap with Marfan syndrome (MFS), osteogenesis imperfecta (OI) and Ehlers-Danlos syndrome (EDS). It

is widely acknowledged that there is little to differentiate between the hypermobility form of Ehlers-Danlos syndrome – previously known as EDS type III and JHS [15]. JHS is accepted as a common clinical entity in musculoskeletal medicine with a prevalence of between 30% - 60% in those presenting to rheumatology or physiotherapy clinics [16-18]. Hypermobility and JHS are assessed by employing the Beighton score [19] and Brighton criteria [20] respectively.

Patients with JHS also report neurophysiological symptoms which include autonomic dysfunction [21,22] pain enhancement [23] and impaired joint proprioception [24]. It is suggested that overuse injuries and predisposition to musculoskeletal pain might be as a result of impaired motor control and biomechanical dysfunction. Biomechanical dysfunction and impaired motor control observed as poorly coordinated movements impacting on function are attributed to developmental coordination disorder (DCD) [25].

DCD is an inherited neurodevelopmental disorder [26] presenting with a variety of functional difficulties [27]. Described in the literature utilising the terms apraxia [28]; clumsy child syndrome [29]; perceptual

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motor dysfunction [30]; dyspraxia [31] the term DCD was endorsed at an international consensus meeting in London, Canada [32]. DCD is referred to in the Diagnostic and Statistic Manual for Mental Disorders fourth edition DSM-IV-TR [25] and in the International classification of mental and behavioural disorders (ICD-10) [33] under the term specific developmental disorder of motor function. It is assessed by health professionals by employing checklists and batteries of tests [34,35]. It is well described in children but there is limited literature in relation to adolescents and adults [36,37]. DCD occurs on a continuum and the prevalence range of 1.6%-34% reflects this aspect. This range is further compounded by the different assessment tools and cut-off scores which have been employed in studies [38,39,40,41].

Children with JHS are reported to show similar functional difficulties as children with DCD [27]. These include not only motor control difficulties but difficulties with reading and spelling. Children with DCD are more likely to report musculoskeletal pain and symptoms of autonomic dysfunction than children without DCD [42].

There is anecdotal evidence to suggest that deconditioning is an important feature for patients with JHS [43,44]. Deconditioning occurs as the result of a loss of physical fitness. Physical fitness is associated with both health and skill related attributes. It is suggested that the multisystemic nature of JHS which affects the musculoskeletal and visceral systems may contribute to a reduction in health related physical fitness. Biomechanical dysfunction and motor control impairments observed in some individuals with JHS may be as a result of an association with DCD. This association may further contribute to deconditioning as it affects skill related physical fitness. The aim of this literature review is to examine the evidence relating to an association between joint hypermobility, JHS, motor control impairments, developmental delay and DCD.

## Method

### Search strategy

In this next section the literature search is described and a critique and summary of the relevant articles is presented in the results. An initial search for literature related to both JHS and DCD was carried out using the following data bases: EMBASE, MEDLINE, CINAHL, ASSIA, PsychARTICLES, SPORTDiscus and PsycINFO from 1989 - 2009. The key words used to search each data base included joint hypermobility syndrome (JHS), benign joint hypermobility syndrome (BJHS), hypermobility, and Ehlers-Danlos syndrome (EDS) with

developmental coordination disorder (DCD), dyspraxia, impaired motor development, clumsy child syndrome and coordination. A manual search of the reference lists from each article was conducted. Articles were included for review if they were in English and in peer reviewed journals.

### Quality of the evidence

The quality of the evidence was assessed by employing the evidence grading system described by Greer et al. [45]. The details of which are presented in table 1. The evidence grading system [45] was introduced by the institute of clinical systems improvement (ICSI) in 1996. The aims of the grading system were to provide a method for reaching evidence-based conclusions, for grading the conclusions, the strength of the evidence and to increase the systematic use of evidence. The experience and results of employing this system were presented in 2000 [39].

## Results

### Results of the search

There were five primary research studies retrieved from the literature searches and subsequently evaluated (See table 1). Two studies which assessed motor development impairments and joint hypermobility were longitudinal observational comparison studies in infants and young children. The quality of these studies were categorised as level I. They took place in clinical populations in Israel, participants that were assessed at follow up were randomized [46,47]. A third study the quality of which was categorised as a level III was a cross sectional case comparison observational study. This study compared joint hypermobility and motor development in school aged children in the first and second grades of three elementary schools and the first grade of a special education school in Israel [48]. The fourth study was a retrospective observational study and categorised as a level III study. This study compared joint hypermobility and motor development in infants and children. This was carried out in a clinical population in the Netherlands [49]. The fifth study was a survey case comparison study the quality of which was categorised as level III. This study compared functional impairments in children with a diagnosis of JHS and those with a diagnosis of DCD. The children with JHS were all members of the Hypermobility Association, which is a national organisation in the United Kingdom. The children with DCD were from a specialist neurodevelopmental clinic [27].

Author, year and country of study [reference]	Jaffe et al 1988, Israel [40]	Tirosh et al 1991, Israel [41]	Davidovitch et al 1994, Israel [42]	Englebert et al, Netherlands [43]	Kirby et al 2005, United Kingdom [27]
<b>Study, design type</b>	A	A	B	C	C
<b>Quality of individual research reports based on answers to the following (Y = Yes; N = No)</b>					
Inclusion exclusion criteria well defined and adhered to	Y	Y	N	Y	Y
No serious questions of bias	Y	Y	Y	N	N
Statistically significant and clinically important effect	Y	Y	N	N	N
Results generalizable to other populations	Y	Y	N/Y	Y	N/Y
Characteristics of a well-designed study clearly addressed	Y	Y	N	N	Y
Overall quality of study	+	+	∅	∅	∅
Conclusion Grade (Level)	I	I	III	III	III

**Study Design type:** A = randomised, controlled trial; B = Cohort study; C = nonrandomized trial with concurrent or historical controls, case-control study, diagnostic test with sensitivity and specificity, population-based descriptive study; D = cross-sectional study, case series, case report

**Quality of study:** If a study has 2 or more YES it may be designated a (+); If a study has 2 or more NO it may be designated a (-); If answers pertaining to (+) or (-) do not indicate the report is exceptionally strong or weak then the report will be graded as neutral ∅

**Conclusion grades:** Level I – level III and Grade not assignable. Conclusion supported by: Good evidence (Level I); Fair evidence (Level II); Limited evidence (Level III). Grade not assignable where there is no evidence to support or refute conclusion

**Table 1:** Results of primary research report quality categories employing an evidence grading system [39].

## Participants

Participants were restricted to infants and children under the age of 12 years in all the studies [27,46-49]. Sex distribution of children was only reported in one study [48]. This is a limitation of these studies as hypermobility and JHS tend to be more prevalent in females while DCD is reported more frequently in males. The mean ages of the infants and children as a whole group or within groups was not reported in three studies [46-48]. The mean age of the 72 children in the study conducted in the Netherlands was reported as 5.4 years (SD 2.5, range 1.3 – 11.6) [49]. The study consisted of two defined groups; infants under 2.5 years (16 participants) and children aged 4 – 12 years (56 participants). The mean ages, SD and age ranges of these groups were not reported. In the study that investigated functional difficulties between two populations of children with either JHS or DCD [27]. There were 68 children with JHS (9 years and 7 months) and 58 children with DCD (8 years). This study omitted data relating to SD and age ranges. One longitudinal study started with 715 infants aged 8 – 14 months. Of these 197 randomly selected infants were followed up 6 months later [46]. In the following longitudinal study which followed up participants from a previous study [46] a total of 59 children were followed up at between 54 and 60 months and divided equally in three groups. In another study conducted in Israel 320 school children were recruited from first and second grades in three elementary schools and 110 first graders from a special education school data relating to mean age, SD or age ranges was omitted [48].

## Outcome measures

A variety of outcome measures were employed to assess motor function; this reflected in part the different age groups assessed and the location of the study. Children over the age of 4 years were assessed by employing the Movement Assessment Battery for Children (MABC) [34], in the UK and Netherlands. In the Netherlands data was reported for children scoring between the 5<sup>th</sup> and 15<sup>th</sup> percentile and children scoring below the 5<sup>th</sup> percentile [49]. In the UK children were recruited to the study if they scored below the 5<sup>th</sup> percentile of the MABC [27]. While in Israel they were assessed by Tirosh and colleagues [47] by employing the Bruninks-Osteretsky Test of Motor proficiency BOT-MP [35]. Davidovitch and colleagues employed a neurological examination for school children [48]. The Beery-Butencia visual motor integration test (VMI) [50] was reported by two studies [47,48]. One study employed the Hoskins-Squires test [51] for gross motor and reflex development [47]. Infants aged 8-14 months in Israel [52] were assessed by employing the Denver development screening test [52].

A variety of measures were employed to assess hypermobility. The Carter Wilkinson test [53] and two additional sites were assessed in the three studies that recorded hypermobility in Israel [46-48]. In the Netherlands the Bulbena criteria [54] were employed [49]. In the study in the UK the assessment strategy was not documented although all children reported a diagnosis of JHS. In the UK the Beighton score [19] and Brighton criteria [20] are employed for the assessment of JHS in specialist centres [55].

## Sample bias

Sample bias was acknowledged by the authors of two studies. In the study in the Netherlands there were 200 children with generalised hypermobility whose records were reviewed retrospectively. Of the 200 children 72 had had a previous assessment of motor development and there was no data to indicate why a motor assessment had been performed in some children and not others [49]. In the study from the United Kingdom the authors acknowledged a bias in the population

selected with JHS for two reasons [27]. The first reason was that the survey was sent to members of a support organisation this might indicate the members were those with a more severe version of the condition. In addition the response rate was low (16%) again which might indicate that only those with more severe difficulties contributed to filling in the questionnaire.

It is possible there was bias in the population involving children attending a special education school and three elementary schools in Israel [48]. The population observed 1<sup>st</sup> and 2<sup>nd</sup> graders in three elementary schools and only a population of 1<sup>st</sup> graders in the special education school. There was no explanation as to why only one year group were recruited from the special education school. In addition 20 children with joint hypermobility and 20 controls were observed in both the 1<sup>st</sup> and 2<sup>nd</sup> grades of the elementary schools. Only six children with or without joint hypermobility were observed in the special education school. There was no indication of why children were attending the special education school. It is possible that the neurological examination employed in this study was not sensitive enough to assess motor development, because no group with developmental delay was reported [48]. However, this study also employed the VMI test [50] and the results indicated a difference in the VMI scores between children in the elementary schools and those in the special education school [48].

## Joint hypermobility and motor delay or developmental dysfunction

In the study in which 715 infants aged 8 - 14 months were examined it was found that those with three or more hypermobile sites were significantly more likely to have motor delay. Follow up six months later in this study revealed that motor delay had persisted in some infants with joint hypermobility but this was not statistically significant [46]. In a subsequent study of 59 children who were examined at the age of 5 years there was a significant association between motor delay and joint hypermobility compared with the group who were not hypermobile [47]. The results of a study of children attending a special education and elementary schools found there was no association between joint hypermobility and neurodevelopmental dysfunction [48]. In a study of 56 children aged between 4-12 years 25% were found to have developmental delay and 21% at risk of motor delay in addition the mean age of walking for the group was 18 months [49].

## Joint hypermobility syndrome and developmental coordination disorder

There was only one study that investigated the similarities in functional difficulties reported by 68 children with JHS and 58 children with a diagnosis of DCD. This study found that children with JHS reported a range of difficulties that were similar to children with DCD [27].

## Discussion

This review has identified few primary research studies relating to an association between joint hypermobility, JHS and impaired motor development and DCD. This is partly because it is difficult to evaluate the association between motor control impairments and hypermobility. Joint hypermobility in the studies reviewed have been reported by employing a variety of systems which include Carter Wilkinson [53], Beighton [19] and Bulbena [54]. This should not be a cause for concern as they are reported to be highly correlated [54]. More importantly there is a misconception that higher scores recorded by the hypermobility assessments relate to a higher degree of hypermobility. This misconception was apparent in Davidovitch and colleagues' study

[48] in which the relationship between degree of hypermobility and motor delay was assessed. In addition norms for joint hypermobility in the very young age groups have not been defined.

The criteria for Joint hypermobility syndrome have only more recently been described and have not been validated for children. Although in specialist centres JHS is diagnosed in the young population through a clinical examination [56]. A variety of assessment tools were employed to establish motor control impairments or delay and this is because the different assessment tools are validated for different age groups and assess a variety of aspects in relation to motor control impairments.

### Overall completeness and applicability of the evidence

From the studies described for this review it would appear that this topic has not been fully addressed. The evidence suggests this is an area which requires further consideration. In the earlier studies although the literature identified motor control difficulties and delay in motor development these were not referred to as DCD [40-42]. The diagnostic criteria for DCD only appeared in the DSM-III in 1987 [57] and the term DCD was only more formally acknowledged following an international consensus in 1995 [32]. It is only the study by Kirby et al. [27] that highlights similarities in the functional difficulties between children with a diagnosis of DCD and those with a diagnosis of JHS. In addition hypermobility and symptoms in the form of a non inflammatory connective tissue disorder - JHS were only more formally acknowledged with the publication of the Brighton criteria in 2000 [20].

### Limitations in the light of the quality of the evidence

Limitations of this review include difficulty drawing general conclusions about the results of the studies. This was primarily because the studies employed a variety of methodologies. In addition there was evidence of sample bias, inadequate description of the inclusion criteria and description of the participants (mean age and sex). Some of the studies reported significant differences, others only descriptive statistics. Between study comparisons were compounded by the fact that assessment tools were not similar, cut-off scores were not always recorded and methodology of assessment were not always adequately described.

### Other studies considered

Pain and clumsiness were the commonest features reported in a study of children and adolescents aged from 3 -17 years with a diagnosis of JHS attending a tertiary referral hospital [56]. The aim of the study by Adib et al. [53] was to characterise the clinical profile of those with JHS. In addition to a range of musculoskeletal and visceral symptoms the participants reported abnormal gait, fine motor control difficulties and delayed walking. Delayed walking was observed in infants with joint hypermobility by Mintz-Itkin et al. [58] most of whom were walking by 18 months following an intervention program. It might be suggested that intervention programs targeting muscle strength contribute to improving motor performance. Muscle strength was positively associated with motor performance in children with joint hypermobility [59] and significant improvements in proprioception were recorded in adults with JHS following a graded exercise program [60].

Adib et al. [56] suggested that motor impairments observed in children with JHS are likely to be related to the central nervous system and impaired proprioception [56]. Impaired proprioception in adult patients with JHS has previously been reported with anecdotal evidence

to suggest that some patients reported 'clumsiness' in childhood [60]. Impaired proprioception is a common feature in those with DCD [30] and is thought to contribute to delayed motor development [61] and motor competence. Conversely in a study of 8 year old children in Denmark [62] no significant correlation was found between children with joint hypermobility and various tests of motor competence. As previously suggested the number of hypermobile sites does not equate to the degree of hypermobility and therefore this comparison may not be valid. Interestingly Juul-Kristensen et al. [62] also reported on a small number of children with JHS who were no more likely to report impaired motor competence than children without JHS. Just over a third of the children in this study with JHS reported musculoskeletal pain [62]. There was no assessment relating to the association between JHS, musculoskeletal pain and motor competence. It is suggested that pain may be a salient feature in those with both JHS and DCD; this is an area to explore in future studies.

Ferrell et al. [60] suggested that children with hypermobility, delayed motor development and impaired proprioception may go on to develop pain in later life. Pain has been reported as a significant feature for children with a diagnosis of DCD [42], but this is not a feature commonly reported in the literature pertaining to DCD. There is emerging evidence that children with DCD continue to report motor control and coordination difficulties in adulthood [36,37], however, this has not been linked to musculoskeletal symptoms, pain or JHS.

### Conclusion

The aim of this review was to explore the literature in relation to identifying a possible association between joint hypermobility, JHS and motor delay and DCD. There was limited literature relating to an association between joint hypermobility, JHS, impaired motor control, motor delay and DCD in infants and children and there was no literature to describe these features in adults. It appears that this topic has not been comprehensively addressed. There is evidence that joint hypermobility is associated with motor delay and motor control impairments in young children but this is not conclusive. The discrepancies in the results may be as a result of methodological differences. There was evidence of similarities in functional difficulties reported by children with a diagnosis of DCD and children with a diagnosis of JHS.

It is suggested that the biomechanical dysfunction and motor control impairments observed in some individuals with JHS who report pain may be as a result of an association with DCD. It is also suggested that this association may contribute to reduced physical activity and deconditioning which further contribute to biomechanical dysfunction and pain. As previously suggested [62] there is a requirement to explore the long term nature of an association between JHS and DCD and chronic pain. In addition there is a requirement to explore whether patients with JHS and DCD report reduced physical activity. Understanding the relationship between JHS and DCD and the manifestations will enable professionals to recognise, acknowledge and address features of these conditions that continue to be troublesome through life.

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