Is Motor Performance in 5.5-Year-Old Children Associated with the Presence of Generalized Joint Hypermobility?

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Objective To determine the prevalence of generalized joint hypermobility (GJH) in Dutch children aged 5.5 years, and to examine the association between GJH and motor performance and development over time.

Study design A prospective cohort of 249 children was recruited. GJH was assessed with the Beighton test at age 5.5 years. Motor performance was evaluated at age 2.0 years using the Bayley Scales of Infant Development, Second Edition and at age 5.5 years using the Movement Assessment Battery for Children–Second Edition (subscore categories: manual dexterity, aiming and catching, and static and dynamic balance).

Results In 249 children, the prevalence of GJH, defined by the Beighton test score, was 34.1% for a score ≥4, 22.5% for a score ≥5, and 16.5% for a score ≥6. No significant association was found between GJH and total motor performance. Manual dexterity in girls (Beighton score ≥4) was positively associated with higher level of motor performance (β [SE] = 0.38 [0.17]; P = .028), ranging from +0.04 SD to +0.72 SD, even after correction for covariates. A significant interaction between GJH and body mass index (BMI) growth was found, indicating that the effect of GJH on the rate of development of motor performance declines with increasing BMI growth (β = 0.05 [0.02]; P = .031).

Conclusion In this healthy pediatric cohort, GJH was present in one-third of the sample, and no significant association was found between GJH and total motor performance. The effect of GJH on the rate of development of motor performance appears to decline with increasing BMI growth. Longitudinal prospective studies are recommended to detect influences of GJH on motor performance over time, as well as the influence of body composition and Beighton cutoff points. *(J Pediatr 2015;167:694-701)*.

Generalized joint hypermobility (GJH) is common in children. When arthralgia in more than 4 joints is present for longer than 3 months without any signs of rheumatic, neurologic, skeletal, or metabolic disease, hypermobility syndrome (HMS) can be diagnosed.[1] The presence of GJH in children is commonly detected using the Beighton test. This measure is considered the gold standard from infancy to old age.[2-5] Use of the Beighton test is controversial, however, owing to a lack of standardization in children, as well as a lack of age-, sex-, and ethnicity-specific cutoff values.[2,4,6]

The prevalence of GJH is unclear, with extensive variation reported in the relevant literature. This could be explained by the absence of an international consensus and the lack of operationalization standards, which vary according to differences in study populations, cutoff levels, and actual administration of the Beighton test.[2,6-8] Murray et al[9] reported GJH prevalence ranging between 2% and 55% in various pediatric populations. Recent studies have examined the heterogeneity of GJH in the Caucasian population by comparing the prevalence of GJH at varying cutoff levels and taking age and sex into consideration.[2,4,6,10,11] Differences in cutoff levels also might be explained by the more precise and detailed descriptions of standard operating procedures used for measuring local joint hypermobility and GJH, as first described by Juul-Kristensen et al.[3]

The concurrent validity of the Beighton test in relation to goniometry has been found to be high.[4] Regarding predictive value, one study found that 10-year-old children diagnosed with GJH and musculoskeletal pain using the Beighton test had an increased risk of persistent pain at age 14 years.[12]

Despite the prevalence of GJH in normal populations,[13,14] much remains unknown about its consequences. GJH has been associated with a wide variety of musculoskeletal complaints, including joint pain, dysfunction of various organ systems (eg, blood vessels and skin), and psychosocial problems.[2,6,15-18] GJH is associated with an increased incidence of motor delay in infancy, with

AICC	Akaike information criterion
BMI	Body mass index
BSID-II	Bayley Scales of Infant Development, Second Edition
GJH	Generalized joint hypermobility
HMS	Hypermobility syndrome
MD	Mean difference
Movement ABC-2	Movement Assessment Battery for Children–Second Edition

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catch-up occurring in most children by age 2 years.[19] In one study, however, at age 5 years, motor performance was significantly delayed in children who exhibited joint hypermobility and motor delay at age 18 months.[20] Adib[21] reported clumsiness, poor motor coordination in early childhood, and problems with handwriting tasks. Engelbert et al[22] observed severe delay in motor development in approximately one-third of children with GJH, with no association with the number of hypermobile joints.[22] Juul-Kristensen et al[10] and Remvig et al[2] evaluated the prevalence of GJH (at different cutoff levels) in primary school children at age 8 and 10 years and concluded that motor competence was not delayed.

In the cohort reported here, we previously determined the relationship between joint hypermobility and motor performance with and without developmental coordination disorder, and found a significant negative correlation between joint mobility and motor performance in the children with developmental coordination disorder, but not in the group of typically developing children.[23]

The purpose of the current study was 3-fold: (1) to determine the prevalence of GJH as defined by the Beighton test at various cutoff levels in Dutch children aged 5.5 years; (2) to study the association between GJH and specific issues of motor performance at age 5.5 years; and (3) to evaluate the consequences of GJH on the rate of motor development over time in children aged 2.0 years and 5.5 years.

Methods

We recruited 249 healthy Dutch children (mean age, 5.5 years) from a birth cohort study of 400 healthy term neonates at the General District Hospital Bernhoven at Veghel, The Netherlands. This cohort was part of 2 earlier studies by van Vlimmeren et al,[24,25] in which assessment of motor performance was performed at age 2 and 5.5 years. Assessments were performed by a team of 12 qualified pediatric physiotherapists. The interrater reliability was high, measured before the start of the study (intraclass correlation >0.8). Medical ethical approval was given by the Committee for Medical Ethics at University Medical Center Utrecht, Radboud University Medical Center, and Bernhoven Hospital Veghel, The Netherlands. Written informed consent was obtained from all parents in accordance with the Declaration of Helsinki.

Anthropometry

Demographic data were collected regarding age, sex, height, and weight. Standing height and weight were measured using a standardized method (wearing underwear and bare feet, measured to the nearest 1 cm and 100 g, respectively). Body mass index (BMI) was calculated as weight in kilograms divided by height in meters squared.[26]

GJH According to Beighton Score

To determine the presence of GJH, the protocol published by Smits-Engelsman et al[4] was used. The Beighton test is a reliable[3] and valid instrument for classifying GJH in primary school-aged children.[4] The Beighton test consists of 5 clinical maneuvers and is scored dichotomously (0-1). A total score (ranging from 0 to 9) was derived by summing all maneuvers: (1) bilateral passive apposition of the thumb to the flexor side of the forearm, with positive score if the whole thumb touches the flexor side of the forearm (shoulder 90° flexed, elbow extended, and hand pronated); (2) bilateral passive dorsiflexion of the fifth metacarpophalangeal joint ≥90° (sitting on chair, arm in 80° abduction, elbow flexed 90°, forearm resting on a table, forearm pronated); (3) bilateral passive hyperextension of the elbow ≥10° (sit on chair with shoulder 90° anteflexion, forearm supinated); (4) bilateral passive hyperextension of the knee ≥10° (lying supine with legs supported by a table); and (5) forward flexion of the trunk, with knees straight, so that the palms of the hands rest easily on the floor.[4]

The presence of GJH was used for analysis at 3 cutoff points: Beighton score ≥4 (GJH4), Beighton score ≥5 (GJH5), and Beighton score ≥6 (GJH6). We used these different cutoff points because the correct cutoff point in children is still under discussion (number of positive items, related to sex). When applying GJH7, only 25 (19 girls) children were scored with GJH. When these data were applied to the model, the basic statistical assumptions could not be fulfilled, owing to the variance of the outcomes in relation to the small subsample size.

Motor Performance Tests

Motor performance was determined by age-appropriate test batteries at age 2.0 years by the motor scale of the Bayley Scales of Infant Development, Second Edition (BSID-II)[27] and at age 5.5 years by the Movement Assessment Battery for Children–Second Edition (Movement ABC-2).[28,29] For both the BSID-II and Movement ABC-2, the deviation of normality on the basis of normative values was incorporated into the scores.

In the total cohort, the English version of the BSID-II was used to score motor performance at age 2.0 years.[27] This instrument comprises 3 scales—the mental, motor, and behavior rating scales—but only the motor scale was used for the present study. The motor scale is a highly reliable, valid, and norm-referenced instrument for evaluating the general development of infants aged 1-42 months.[27] This scale consists of 111 items that measure skills related to gross and fine motor control. The items in the motor scale are task items, scored dichotomously. Raw scores were converted into a Psychomotor Developmental Index score, which has a mean (SD) of 100 (15).[27]

The Movement ABC-2 was used to assess motor performance at age 5.5 years. The Movement ABC-2 is a standardized and norm-referenced test, validated for the Dutch population, that aims to classify children aged 3-16 years according to degree of motor performance.[28,29] This test consists of 8 items in each of 3 age groups (3-6 years, 7-10 years, and 11-16 years). These items measure different aspects of motor performance divided into 3 major areas: manual dexterity, aiming and catching, and static and dynamic balance. For each child, the raw item scores were