THE JOURNAL OF PEDIATRICS • www.jpeds.com

ORIGINAL ARTICLES

Joint Laxity in Preschool Children Born Preterm

Domenico M. Romeo, MD, PhD[1], Chiara Velli, PT[1], Simona Lucibello, MD[1], Gloria Ferrantini, MD[1], Giuseppina Leo, MD[1], Claudia Brogna, MD, PhD[1], Francesco Cota, MD[2], Daniela Ricci, MD, PhD[3], Francesca Gallini, MD[2], Costantino Romagnoli, MD[2], Giovanni Vento, MD[2], and Eugenio Mercuri, MD, PhD[1]

Objective To evaluate the prevalence of joint laxity in children born preterm assessed in the first 2 years, the relationship between joint laxity and motor performance at preschool age, and possible changes over time in a subgroup of children followed longitudinally.

Study design The revised scale of Beighton Score was used to evaluate joint laxity in a population of 132 preschool children born preterm between 24 and 32 weeks of gestational age. All were assessed for joint laxity between 12 and 24 months of age. Children also performed the Movement Assessment Battery for Children–Second Edition between the age of 3 years and 6 months and 4 years; the age at onset of independent walking also was recorded.

Results The total Beighton Score ranged between 0 and 8. Twenty percent of the cohort showed joint laxity. No differences related to sex or gestational age were observed. Children born preterm with joint laxity achieved later independent walking and achieved lower scores on Movement Assessment Battery for Children–Second Edition than those without joint laxity. In 76 children born preterm, an assessment for joint laxity was repeated once between 25 and 36 months and again after >36 months. No statistically significant difference was observed between the 3 assessments.

Conclusions The Beighton Score can be used to assess generalized joint laxity in children born preterm. As the presence of joint laxity influenced motor competences, the possibility to early identify these infants in the first 2 years is of interest to benefit from early intervention and potentially improve gross motor skills and coordination. *(J Pediatr 2018;■■:■■-■■).*

Joint hypermobility is defined as greater than normal joint motion in a single joint or generalized.[1] The etiopathogenesis is not clearly known, but it is assumed that both environmental and genetic factors could be involved. Joint hypermobility is a clinical feature of various hereditary diseases of connective tissue, such as Ehlers–Danlos syndrome or Marfan but is more often clinically observed as an isolated phenomenon, defined as asymptomatic hypermobility.[2-4]

The prevalence of asymptomatic hypermobility in children varies from 3% to 30%[2-5] depending on the inclusion criteria. Recent studies focusing on children younger than the age of 5 years reported a prevalence of 7% of hypermobility,[4] defined via a revised version of the Beighton Score, which is designed to quantify joint hypermobility. This value is greater than the prevalence found in most of the studies reporting joint hypermobility in pediatric populations mainly including school-aged children.

Joint laxity often is observed in children born preterm,[6] but no systematic study has been performed to study its prevalence or its possible effect on milestones. Joint laxity refers to a combination of joint hypermobility with muscle extensibility. The aims of the present study were to evaluate the prevalence of joint laxity by using the Beighton Score in children born preterm assessed in the first 2 years of life, study the relationship between the Beighton Score and motor performance at preschool age, and to examine possible changes over time in a subgroup of children followed longitudinally.

Methods

Children were recruited from the Neonatal Unit at the Policlinico Gemelli Hospital in Rome, Italy, from January 2011 to January 2013. Informed parental consent for the study was obtained for all infants. Infants were enrolled consecutively if they were born between 25.0 and 31.9 (<32) weeks of gestational age, as determined from the results of first trimester ultrasound scans. In addition, their cranial ultrasound scanning results were normal or showed only a transient flare or germinal layer hemorrhage during the first 2 postnatal weeks and at term-equivalent age had no parenchymal abnormality or evidence of atrophy (defined as ventricular dilatation with a ventricular index of >14 mm, irregular ventricular margins, widened interhemispheric fissure, or enlarged extracerebral space).[7] Infants were not included if they were still oxygen-dependent at

M-ABC-2	Movement Assessment Battery for Children–Second Edition

From the [1]Pediatric Neurology Unit; [2]Neonatal Intensive Care Unit, Catholic University, Fondazione Policlinico A. Gemelli, Rome, Italy; and [3]National Centre of Services and Research for Prevention of Blindness and Rehabilitation of Visually Impaired, Rome, Italy

The authors declare no conflicts of interest.

0022-3476/$ - see front matter.
https://doi.org10.1016/j.jpeds.2018.02.008

term-equivalent age, had major congenital malformations, had genetic or chromosomal abnormalities, had known metabolic disorders, had congenital infection or any sign of encephalopathy or seizures during the neonatal course, or had greater than stage 2 of retinopathy of prematurity at the time of the assessment.

Assessment of Joint Laxity

Joint laxity was scored according to the revised version of the Beighton Score suitable for preschool-aged children,[4] consisting of the following:

(1) Passive dorsiflexion of the fifth metacarpophalangeal joint. Score is positive if ≥90° (bilateral testing).
(2) Passive hyperextension of the elbow. Score is positive if ≥10° (bilateral testing).
(3) Passive hyperextension of the knee. Score is positive if ≥10° (bilateral testing).
(4) Passive apposition of the thumb to the flexor side of the forearm, while shoulder is 90° flexed, elbow extended, and hand pronated. Score is positive for joint laxity if the whole thumb touches the flexor side of the forearm (bilateral testing).
(5) Passive dorsiflexion of the ankle joint bilaterally. Score is positive when the angle is >30° (bilateral testing).

A plastic 180° goniometer was used to measure the passive bilateral dorsiflexion of the fifth metacarpophalangeal joint and ankle and the passive bilateral hyperextension of the elbow and knee. The 5 items were performed both on the right and the left side, giving a score of 1 if the item detected hypermobility of the joint according to the instruction of each item and 0 if this was not detected. The total score could range from 0 (absence of joint laxity) to 10 (joint laxity on all items on both sides). All the children were assessed between 12 and 24 months of corrected age.

Interobserver Reliability Assessment

Two examiners both experienced in performing neurologic assessments in infants and children performed the assessments. Interobserver reliability was measured in 20 infants born preterm assessed either between 12 and 24 months of corrected age or at preschool age. The maximum difference between the observers was always ≤1 in the whole assessment. The interobserver agreement was 97%.

Perceptual Motor Abilities

All the children were assessed with the Movement Assessment Battery for Children–Second Edition (M-ABC-2)[8] between the age of 3 years and 6 months and 4 years. The M-ABC-2 includes 3 items assessing manual dexterity, 2 assessing aiming and catching, and 3 assessing balance. Individual results can be compared and scored according to available normative data. The test is designed to give a global score, but also subscores for the 3 subscales also can be compared with age-specific normative data. All the assessments were administrated by specifically trained pediatric neurologists. Using the age-specific normative data available from the manual, we scored the results as normal (when above the 15th percentile), borderline (when below the 15th percentile but above the fifth), and abnormal (when below the fifth percentile). We also noted whether the child refused to perform the test or part of it. The results were classified according to the age-appropriate normative findings and plotting the number of normal, borderline, and abnormal results.

Neurologic Assessment

All children were further examined neurologically using the Touwen Infant Neurological Examination.[9,10] This include assessment of muscle tone, balance and coordination, reflexes, and cranial nerves.

The age at onset of independent walking, defined as the time when the infant was able to take 5 consecutive steps without support, also was recorded.[11] Parents were instructed to note the exact date when the infant began walking independently and to take video photography. Regular phone calls (at least every 2 months) were made to the parents after the infants reached 12 months of corrected age to ensure accurate documentation of walking. A neurologic assessment was performed routinely at 12 months and again at 16-18 months of corrected age, as part of our follow-up of infants born preterm. Approval was obtained from the hospital's ethics committee.

Statistical Analyses

The characteristics of participants are described via mean ± SD. Differences between infants born preterm with and without joint laxity for sex were performed with the Fisher exact test; gestational age and birth weight differences were assessed by the Mann–Whitney *U* test or *t* test as appropriate.

Each item of the Beighton Score and total score was presented as a number and percentage. The frequency distribution of each item and the total scores were calculated in the entire cohort. A cut-off score >4 was used to identify joint laxity according to reference data previously published.[4]

The patients were subdivided in 3 groups according to their gestational age (24-28, 29-30, and 31-32 completed weeks). A further division was made according to the age the children were assessed (≤24 months of corrected age, 25-36 months, >36 months). Mixed model analysis was used to assess the effect of time of observation in the entire cohort and of time of observation and gestational age, considering the interaction between the main independent variables. Pearson correlation was used to test the correlation between Beighton Scores assessed at <24 months of corrected age and age of independent walking and the results of the M-ABC-2 test. The effect of joint laxity on motor development was further investigated by means of linear regression, with each independent variable tested against the Beighton Score, alone and in a model with gestational age to account for confounding.

The level of significance was set at $P < .05$. Data were analyzed with Stata software, version 13 statistical package (StataCorp LP, College Station, Texas).

FLA 5.5.0 DTD ■ YMPD9798_proof ■ April 9, 2018