

Assessing Joint Hypermobility, Proprioception, and Developmental Functioning in Toddlers Born Preterm

Bilge Nur Yardımcı-Lokmanoğlu¹, Akmer Mutlu¹

Hacettepe University Faculty of Physical Therapy and Rehabilitation, Developmental and Early Physiotherapy Unit, Ankara, Turkey

What is already known on this topic?

- Joint hypermobility is common in the children born preterm. It was known that joint hypermobility could affect proprioception and motor development in children, and all of them related to each other; however, there is limited research on toddlers.

What this study adds on this topic?

- A total of 26.79% of the toddlers born preterm had joint hypermobility, and the most frequently occurring joint hypermobility item was "passive dorsiflexion of the ankle." It was seen that there were no differences in the proprioception findings, including both cooperation rate and success rate, and developmental functioning outcomes, including cognitive, language, and motor development, according to joint hypermobility in toddlers born preterm. Additionally, there was no relationship between assessment findings as pairs.

Corresponding author:

Bilge Nur Yardımcı-Lokmanoğlu
✉ bilgenuryardimci@hacettepe.edu.tr, bilgenuryardimci@gmail.com

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ABSTRACT

Objective: The aims of this study were to explore (i) the joint hypermobility, proprioception, and developmental functioning in toddlers born preterm, (ii) differences in the proprioception and developmental functioning between toddlers with and without joint hypermobility, and (iii) the relationship between them.

Materials and Methods: One hundred twelve toddlers born preterm between 24 and 42 months of age were included in this observational study. Beighton Score for joint hypermobility assessment and the Bayley Scales of Infant and Toddler Development, Third Edition (Bayley-III) for developmental functioning were applied in all toddlers born preterm; however, proprioception assessment could be applied on 55 of 112 (49.11%) toddlers born preterm due to lack of cooperation.

Results: Of 112 toddlers, 30 (26.79%) had joint hypermobility. There were no differences in cooperation rate ($P = .629$) and success rate ($P = .887$) in the proprioception assessment between toddlers with and without joint hypermobility (55 toddlers born preterm), which is similar to the cognitive domain ($P = .430$), language domain ($P = .062$), and motor domain ($P = .619$) in the Bayley-III. Additionally, none of them were related to each other ($P > .05$).

Conclusion: Our study findings showed that joint hypermobility has no effect on proprioception and motor development in toddlers born preterm between 24 and 42 months of age, and there is no relationship between them. The possibility of these results might be that movement repetition and not only proprioception but also other sensory systems could be important in this early period of life.

Keywords: Developmental functioning, joint hypermobility, motor skills, proprioception, toddlers

INTRODUCTION

Preterm birth is defined as births before 37 completed weeks of gestation,¹ and which increases the risk of health problems, including morbidities in the early period of life and developmental problems in the long term.^{2,3} When studies on long-term developmental outcomes were examined, infants born preterm had a higher risk of cognitive problems, language problems, and sensory processing difficulties, in addition to motor problems.⁴⁻⁶ Williams et al⁴ reported that motor impairment was reported in 40.5% for mild-moderate impairment and 19% for moderate impairment in the children born preterm who were not diagnosed with cerebral palsy. Romeo et al⁷ also found that low motor performance was higher in children born preterm who had joint hypermobility than children born preterm without joint hypermobility, another important finding was that these children later achieved independent walking.

Joint hypermobility is defined as a higher than normal range of movement in a single joint or generalized^{8,9} and is called asymptomatic hypermobility when it is observed as an isolated

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phenomenon from musculoskeletal symptoms, such as pain or soft-tissue rheumatism.¹⁰ The prevalence of asymptomatic hypermobility in the children until 20 years of age was found to range from 1.8% to 64.6% in several studies, depending on inclusion criteria such as Beighton total score,^{11–14} and prevalence decreased with age.¹⁴ Furthermore, Romeo et al⁷ also reported 20% joint hypermobility in the preschool children born preterm.

Although joint hypermobility might be an advantage in some sports, such as ballet,¹⁵ studies reported that joint hypermobility was associated with poor proprioception sense in the healthy individuals and children,^{16–18} as well as gross motor developmental delay in the children born preterm.⁷ Proprioception is complex system that derives from mechanoreceptors in the muscle, joint capsule, tendon, ligaments, and cutaneous tactile receptors,¹⁶ and this system has an important role in motor control, motor coordination, and motor planning.¹⁹ Jiang et al²⁰ reported that proprioception was also associated with gross motor development in children between 3 and 6 years of age. In other words, motor development might be affected by not only joint hypermobility but also proprioception.^{7,20}

Motor development is described as adaptive, which is also affected by environment and experience, and increasing gradually complex movement sequences.²¹ Previous studies showed that children with risk factors, such as preterm birth, were at risk for motor developmental delay.^{22,23} Developmental functioning, including the language and cognitive development, was related to motor development.^{24,25}

Interestingly, although differences which are caused by joint hypermobility are seen in the childhood, findings explored that there were no differences in motor performance and physical activity level in later age like adolescents with and without joint hypermobility.²⁶ Therefore, it is important to examine the joint hypermobility, proprioception, and developmental functioning in toddlers born preterm and compare the proprioception and developmental functioning results by separating the toddlers born preterm with and without joint hypermobility.

This study aimed to (i) investigate the joint hypermobility, proprioception, and developmental functioning in toddlers born preterm, (ii) compare proprioception and developmental functioning between toddlers born preterm with and without joint hypermobility, and (iii) explore the relationship among joint hypermobility and proprioception, joint hypermobility and developmental functioning, and proprioception and developmental functioning.

MATERIALS AND METHODS

Study Design and Participants

One hundred twelve toddlers born preterm, admitted to the Developmental and Early Physiotherapy Unit, Faculty of Physical Therapy and Rehabilitation, Hacettepe University, Ankara, Turkey, were included in this cross-sectional study between July and October 2022. The inclusion criteria were: (i) born preterm, (ii) be between 24 and 42 months of age, and (iii) not having neurologic, genetic, or metabolic disorder. Infants who had any risk factor, such as preterm birth, small for gestational age, or intracranial hemorrhage, are followed by

both our unit and different department after discharge from the neonatal intensive care unit until about school age. Among these, infants who met the inclusion criteria were invited in this study. Post hoc power calculation for the differences in the motor development of the Bayley Scales of Infant and Toddler Development, Third Edition (Bayley-III) between groups with and without joint hypermobility showed that the study had higher than 99% power using G*Power 3.1.

Written informed consent was obtained from the parents of all infants. This study was approved by the Non-interventional Clinical Research Ethics Board, Hacettepe University (GO 22/675).

Measurements

The following measurement methods were performed on the same day by 2 experienced physiotherapists who have been working in pediatrics field in our unit, which was designed for children and made them feel comfortable.

Joint Hypermobility Assessment

The revised version of the Beighton Score was applied for assessing the joint hypermobility.^{7,12} It comprises the following 5 subcategories (bilateral testing):

1. Passive dorsiflexion of the fifth metacarpophalangeal joint. Score is positive if $\geq 90^\circ$,
2. Passive hyperextension of the elbow. Score is positive if $\geq 10^\circ$,
3. Passive hyperextension of the knee. Score is positive if $\geq 10^\circ$,
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 if the whole thumb touches the flexor side of the forearm, and
5. Passive dorsiflexion of the ankle joint bilaterally. Score is positive when the angle is $>30^\circ$.^{7,12}

All items were applied both right and left side and took only 5–10 minutes. If the item determines as positive, 1 point was given.^{7,12} The total score ranged from 0 (absence of joint hypermobility) to 10 (joint hypermobility on all items bilaterally).^{7,12} A cut-off score ≥ 4 was used to identify joint laxity.²⁷

Proprioception Assessment

Joint position sense testing was performed according to Gray et al.²⁸ First, the examiner put a sticker on finger of toddler while playing with the toddler with eyes open. The toddler was allowed take sticker or play with it. After this process, the examiner put again a sticker on finger of toddler, but this time toddlers' eyes were closed with parent's hand, and the toddler's arm was moved by the examiner. The toddler was asked to find the sticker with their free hand.²⁸ The test was repeated at least 3 times upper and lower extremities both right and left side if a toddler cooperated (cooperation rate $>0\%$). The whole test process took around 10–20 minutes depending on the cooperation level of the toddlers born preterm. It has been seen that proprioception assessment is more difficult to perform in these group because of cooperation problems due to nature of this age group, especially as the age gets younger. For this reason, we could not apply proprioception assessment to some of toddlers born preterm (50.89%).

Cooperation rate (total hits+total misses/total trials), and success rate (total hits/total hits+total misses) were calculated for each toddler born preterm.²⁸ Additionally, if toddler born preterm hits the sticker less than 50% (cooperation rate > 0%), this test is called abnormal test.

Developmental Functioning Assessment

All toddlers born preterm were examined for developmental functioning using the Bayley-III between 24 and 42 months of age by certified assessor. The composite scores for each domain of Bayley-III were calculated. The Bayley-III took approximately 60-90 minutes to complete, although it depends on the age of toddlers.

The Bayley-III scale is one of the most widely used for assessing the developmental functioning in the early period of life. It consists of the 3 domains: cognitive, language (receptive and expressive), and motor (fine and gross).²⁹ The raw score of each 3 domains is converted into a scaled score (a mean of 10 and a SD of 3), and then composite scores (a mean of 100 and a SD of 15) are calculated for cognitive, language, and motor scales.²⁹ A composite index score below 70 (i.e., <2 SD) in these 3 domains indicates severe developmental delay.²⁹

Statistical analysis

The Statistical Package for Social Sciences Software for Macintosh, version 25.0 (IBM corp., Armonk, NY, USA) was performed for statistical analysis. The normal distribution of the variables was evaluated using analytical methods (e.g., Kolmogorov-Smirnov test) and visual (e.g., histograms, probability plots) to determine whether or not they are normally distributed. Nonparametric tests were conducted when the variables were not normally distributed. Descriptive analyses were presented using mean ± SD and median (min-max) for continuous variables and using n (%) for nominal variables. The assessment results in the between groups were compared using the Mann-Whitney U-test for continuous variables (e.g., success rate of proprioception assessment). A P-value of less than .05 was considered statistically significant for all tests. For the analyses of relationships, we used the Spearman correlation coefficient (2-tailed). The value of the correlation coefficient was classified as 0.90 to 1.00 very high correlation, 0.70 to 0.89

high correlation, 0.50 to 0.69 moderate correlation, 0.30 to 0.49 low correlation, and 0.00 to 0.29 little if any correlation.³⁰

RESULTS

The clinical characteristics of 112 toddlers born preterm are presented in Table 1.

Joint Hypermobility Assessment

Of 112 toddlers born preterm, 30 (26.79%) were found to have joint hypermobility, the total score suggested joint hypermobility (total score ≥4). The total Beighton score ranged between 0 and 10 (mean 2.87 ± 1.92) in all toddlers born preterm. The most frequently occurring joint hypermobility' item was "passive dorsiflexion of the ankle" (95%). "Passive dorsiflexion of the fifth metacarpophalangeal joint" (8%), "passive hyperextension of the elbow" (13.4%), "passive hyperextension of the knee" (9%), and "passive apposition of the thumb to the flexor side of the forearm" (16%) were observed bilaterally in less than half of them. Score 0 was present in 4 (3.57%) toddlers born preterm.

Proprioception Assessment Findings

Proprioception assessment was performed in 55 of 112 (49.11%) toddlers born preterm, while 57 (50.89%) toddlers born preterm did not cooperate (cooperation rate = 0). The cooperation rate ranged between 0.375 and 1 (mean 0.96 ± 0.13), while the

Table 1. Clinical Characteristics of Toddlers Born Preterm

All Toddlers Born Preterm (n = 112)		
Female	n (%)	57 (50.9)
Birth weight, grams	Mean ± SD	1751.63 ± 647.03
	Median (min-max)	1750 (420-4330)
Gestational age, weeks	Mean ± SD	31.83 ± 3.24
	Median (min-max)	33 (23-36)
Assessment age at toddler, months	Mean ± SD	30.61 ± 4.97
	Median (min-max)	29.5 (24-42)
Height, cm	Mean ± SD	89.13 ± 5.51
	Median (min-max)	89 (77-105)
Weight, kg	Mean ± SD	12.82 ± 2.12
	Median (min-max)	12.5 (9-18)

Table 2. Bayley-III Composite Score and Proprioception Assessment in Toddlers Born Preterm

		Toddlers With Joint Hypermobility (n = 18)	Toddlers Without Joint Hypermobility (n = 37)	P ^a
Proprioception assessment (n = 55)				
Cooperation rate	Mean ± SD	0.961 ± 0.15	0.960 ± 0.12	.629
	Median (min-max)	1 (0.375-1)	1 (0.375-1)	
Success rate	Mean ± SD	0.946 ± 0.11	0.945 ± 0.12	.887
	Median (min-max)	1 (0.583-1)	1 (0.400-1)	
Bayley-III composite score (n = 112)		(n = 30)	(n = 82)	
Cognitive domain	Mean ± SD	106.67 ± 19.09	103.90 ± 18.05	.430
	Median (min-max)	105 (70-145)	100 (60-145)	
Language domain	Mean ± SD	105.77 ± 17.43	99.31 ± 17.28	.062
	Median (min-max)	106 (68-150)	97 (65-150)	
Motor domain	Mean ± SD	95.03 ± 15.88	93.63 ± 12.78	.619
	Median (min-max)	97 (61-136)	94 (58-130)	

^aMann-Whitney U-test. P < .05 is statistically significant.
 Bayley-III, Bayley Scales of Infant and Toddler Development, Third Edition.

success rate ranged between 0.40 and 1 (mean 0.95 ± 0.12) in 55 toddlers born preterm. The number of abnormal tests was only 1.

Developmental Functioning Outcomes

Bayley-III was applied on all toddlers born preterm. The Bayley-III composite score ranged between 60 and 145 (mean 104.64 ± 18.29) in the cognitive domain, 65 and 150 (101.04 ± 17.48) in the language domain for, and 58 and 136 (94.01 ± 13.62) in the motor domain for all toddlers born preterm.

Differences in the Proprioception Assessment Findings and Developmental Functioning Outcomes According to Joint Hypermobility

There were no differences in the cooperation rate and success rate between toddlers born preterm with and without joint hypermobility ($P = .629$, and $P = .887$, respectively) (Table 2).

There were no differences on the cognitive domain, language domain, and motor domain between toddlers born preterm with and without joint hypermobility ($P = .430$, $P = .062$, and $P = .619$, respectively) (Table 2).

The Relationship of the Findings of Assessment Methods in Pairs

There was no relationship among joint hypermobility and proprioception, joint hypermobility and developmental functioning, and proprioception, and developmental functioning in the toddlers born preterm (Table 3).

DISCUSSION

This study demonstrated that the frequency of the joint hypermobility was 26.79% in the toddlers born preterm. Other studies reported a wide range of prevalence of the

joint hypermobility between 7% and 64.6% in the preschool children.^{7,12,13} Furthermore, there were no differences in the proprioception findings and development functioning outcomes between toddlers born preterm with and without joint hypermobility, as well as no relationship among joint hypermobility and proprioception, joint hypermobility and developmental functioning, and proprioception and developmental functioning.

Interestingly, the findings of the review conducted by Smith et al³¹ reported that individuals with joint hypermobility had poor lower limb proprioception, although upper limb proprioception results were unclear. Fatoye et al¹⁸ also revealed that children with joint hypermobility aged 8-15 years had poorer knee proprioception than children without joint hypermobility, which is similar to the results in adults reported by Hall et al.¹⁶ However, we did not find any differences in the proprioception between the toddlers born preterm with and without joint hypermobility, as well as the relationship between proprioception and joint hypermobility. The first possible explanation for this might be damage to the receptors due to abnormal joint biomechanics in the long term, which results from joint hypermobility.^{18,32} On the other hand, our method of proprioception assessment, which is applied with a sticker, might be affected by touch sense. Hillier et al³³ reported that some test methods might be controversial for proprioception due to touch sense.

In addition to proprioception, motor development might also be affected by joint hypermobility.⁷ It has been reported that children born preterm with joint hypermobility was able to achieve independent walking in upcoming times.⁷ Recently, Lamari et al³⁴ also found that the higher total Beighton scores showed a tendency to not crawl or crawl differently, delayed ambulation, and impaired school performance. However, several previous studies showed that there was no relationship between the motor performance and joint hypermobility by de Boer et al³⁵ and Engelbert et al.³⁶ The findings of this study provide evidence that developmental functioning, such as motor, language, or cognitive development, did not differ in the toddlers born preterm with and without joint hypermobility and not related to joint hypermobility. The reason of this could be that motor development is not only dependent upon joint hypermobility but also on other accompanying minor problems such as developmental coordination disorder. Jelsma et al³⁷ revealed that motor performance and joint mobility are not related in the healthy children, while motor performance is related to joint mobility in the children with developmental coordination disorder. Furthermore, Romeo et al³⁸ recently reported that there was a higher incidence of motor delay or developmental coordination disorder in children with joint hypermobility.

Proprioception and motor development, which are associated with joint hypermobility,^{7,16-18} were also related to each other.²⁰ Additionally, it was reported that angle error of knee proprioception sense decreased with aging between 3 and 6 years of age.²⁰ Furthermore, some studies previously reported proprioceptive dysfunction related to poor motor performance in children who had atypical development.^{39,40} On the other hand, proprioception comprises the sense of joint position, movement,

Table 3. Examination of the Relationships Between the Findings as Pairwise

	<i>P</i>	<i>r</i>
Joint Hypermobility		
Proprioception assessment (n = 55)		
Cooperation rate	.365	0.125
Success rate	.936	-0.011
Bayley-III (n = 112)		
Cognitive domain	.277	0.104
Language domain	.077	0.168
Motor domain	.358	0.088
Proprioception assessment (n = 55)		
Cooperation rate		
Bayley-III		
Cognitive domain	.582	0.076
Language domain	.798	0.035
Motor domain	.117	0.214
Success rate		
Bayley-III		
Cognitive domain	.420	0.111
Language domain	.517	0.089
Motor domain	.452	0.103

Spearman correlation coefficient. $P < .05$ is statistically significant.
 Bayley-III, Bayley Scales of Infant and Toddler Development, Third Edition.

and force⁴¹⁻⁴³ and plays an important role in motor development together with tactile sense and vestibular sense.^{44,45} The information from proprioception, tactile, and visual sense is required to enable the children to learn a new skill.⁴⁶ Those findings, which were reflected by the present study, demonstrated that not only proprioception but also other sensory systems, especially tactile, visual, or vestibular, might be important for developing new motor skills.

The limitation of this study was that proprioception assessment demands high levels of attention and applies with eyes closed in toddlers which becomes problematic for some toddlers, especially for lower ages.

CONCLUSION

This study demonstrated the results of the joint hypermobility, proprioception, and developmental functioning together in toddlers born preterm between 24 and 42 months of age. There were no differences in proprioception and developmental functioning between toddlers born preterm with and without joint hypermobility and no relationship between them. However, toddlers born preterm might have the risk of joint hypermobility, reduced proprioception acuity, and poor developmental functioning; therefore, detailed assessment may help us to understand the problems in the clinic. On the other hand, the differences might increase with the repetition of movement as getting older. For this reason, results of more long-term follow-up assessment in these toddlers would help us to clarify this. Additionally, along with proprioception, other senses might be important for motor development, and it is suggested that the assessment of other sensory systems should be performed in this period of life in future studies.

Ethics Committee Approval: This study was approved by Ethics Committee of Hacettepe University (Approval No: GO 22/675, Date: 05.07.2022).

Informed Consent: Written informed consent was obtained from all parents of all infants who were included in this study.

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