The Validity of the Gross Motor Function Measure for Use in Extremely Preterm Infants

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Purpose To examine the validity of the Gross Motor Function Measure (GMFM) in assessing gross motor function (GMF) of extremely preterm infants relative to the Alberta Infant Motor Scale (AIMS). ***Methods*** The retrospective study included 60 extremely preterm infants born at <28 weeks gestational who were assessed for gross motor function using GMFM-88 and AIMS at 6 and 12 months of corrected age (CA). The correlations between the AIMS raw scores and the total scores of both GMFM versions at 6 and 12 months CA were assessed. ***Results*** GMFM-66 and GMFM-88 total scores showed moderate to strong correlation with AIMS ($p < 0.001$). GMFM-66 and GMFM-88 scores were significantly different between infants who received AIMS scores above and below the 10th percentile ($p < 0.05$). ***Conclusions*** The GMFM had good validity in assessing GMF in extremely preterm infants at 6 and 12 months CA.

Key Words Extremely preterm infant, Validity, Motor skills, Child development, Gross motor function

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Received date 06 Dec 2021
Revised date 03 Feb 2022
Accept date 08 Feb 2022

Ⅰ. Introduction

Over the last few decades, the mortality rate among preterm infants has decreased, and the survival of extremely preterm infants born before 28 weeks of gestational age (GA) has greatly improved due to advances in perinatal and neonatal intensive care.[1,2] However, infants born extremely preterm are at high risk for of neurodevelopmental delay compared to the general population because of the immaturity of all systems.[3,4,5] Many studies have described the problems with gross motor development in preterm children.[6,7] Problems in gross motor development during the early years of life in preterm infants have a high predictive value for long-term motor problems.[8] Impairment of gross motor function is also an important risk factor for cognitive problems and learning disabilities in later life for preterm children.[9,10] Therefore, early intervention for high-risk extremely preterm infants has attracted an increasing amount of attention.[11]

Assessment of gross motor function is an important component of early interventions for extremely preterm infants who survive. It is recommended that physicians examine the development process of extremely preterm children regularly from early infancy to late childhood. An overview of the developmental trajectory can provide a clearer picture of childhood developmental disorders. There are several standardized measures of early motor development of preterm infants that have been used clinically or in research; these measures include the Test of Infant Motor Performance,[12] Prechtl's Assessment of General Movements,[12] and the Alberta Infant Motor Scale (AIMS).[13,14] However, these validated assessment scales for evaluating gross motor function in infants have a major limitation of short applicatory periods. The AIMS is an investigated tool for measurement gross motor maturation of infant from birth to the age of independent walking. It is known as useful tool because the procedures can administer noninvasively and can be completed in a short amount of time, but an infant who can independently walk can no longer

http:dx.doi.org/10.17817/2022.02.03.1111705

have their gross motor function assessed using the AIMS.[14]

The Gross Motor Function Measure (GMFM) is the most widely used tool for assessing gross motor function in children with cerebral palsy (CP).[15,16,17,18] It was developed for assessing changes in gross motor function rather than identifying disease or predicting future function.[18,19] The GMFM is appropriate for children whose motor skills are at or below those of a 5-year-old child without any motor disability.[15] It is thought that this type of assessment tool, focusing on longitudinal changes in motor function, is suitable for evaluating extremely preterm infants as early as in infancy; this is because a differential diagnosis of CP or other developmental delays is quite difficult.

However, as far as we know, the validity of the GMFM has not been demonstrated for extremely preterm infants in the medical literature. Therefore, validation of the GMFM is indispensable for extending application of the GMFM to all extremely preterm infants, irrespective of a diagnosis of CP. Therefore, the main objective of this study was to validate the GMFM-88 and GMFM-66 for use with extremely preterm infants before one year of age relative to AIMS, the other widely used standardized measurement tool of motor development. A second aim was to suggest reference GMFM scores for detecting delayed motor development in extremely preterm infants.

II. Material and Methods

The GMFM and AIMS were components of a high-risk infant follow-up program in our medical center. This study was performed through a retrospective chart review of prospectively collected data. The infants were assessed with the GMFM-88 and the AIMS on the same day at 6 months (± 2 weeks) and 12 months (± 2 weeks) of corrected age (CA) by two trained pediatric physical therapists. The GMFM-66 scores were determined with the Gross Motor Ability Measure at the time of review. Medical records were reviewed by a pediatric physiatrist to collect the health history of the infants. This study was approved by the local institutional review board.

1. Subjects

Our inclusion criteria were (1) extremely preterm infants who were born before 28 weeks of GA and (2) who were assessed for gross motor function using GMFM-88 and AIMS at 6 and 12 months CA and (3) were followed up lately at ≥72 months of CA. Infants who were diagnosed with chromosomal anomalies, metabolic disease, or any other congenital anomaly, such as serious congenital heart disease were excluded. Whether the infant was diagnosed CP or receiving physical therapy was not considered.

2. Instruments

1) Gross Motor Function Measure

The GMFM-88 and GMFM-66 are validated tools for assessing gross motor function in children with CP.[16,17,18] The original published GMFM had 88 items, subdivided into five dimensions: Lying and Rolling; Sitting; Crawling and Kneeling; Standing; and Walking, Running, and Jumping. Items are scored on a 4-point scale. Scores for each dimension are expressed as a percentage of the maximum score for that dimension. A total score is obtained by adding the percent scores for each dimension and dividing by 5. The more recent and short version of the GMFM, GMFM-66, has only 66 items and can be used to calculate only total scores and not dimension scores.[20] The GMFM-66 has a hierarchical structure with interval scaling that, based on a Rasch analysis, should improve the scoring, interpretation, and overall clinical utility relative to the GMFM-88. GMFM-66 scores were calculated with the Gross Motor Ability Estimator using GMFM-88 scores at the time of chart review. In 5 infants, GMFM was not performed at 12 months of GA.

2) Alberta Infant Motor Scale

The AIMS was chosen as a criterion measure to assess the validity of two versions of GMFM. This is because it is the most commonly used measure of motor development in early intervention programs in our country and it was a standardized assessment tool for preterm infants.[21,22,23] The AIMS is a scale for assessing

gross motor function in infants from birth to 18 months or when a child is able to walk independently. It was validated not only for preterm infants, but also for other infants at risk for motor delays.[23,24] The raw score of the AIMS was used as a basis for comparison. In addition, the 10th percentile of the AIMS score was assumed to indicate abnormal development at each age.

3. Data Analysis

All statistical analyses were performed with SPSS statistical software, version 19.0 (IBM Corporation, New York, USA). Data were reported as median (minimum, maximum, and range) because they lacked normal distribution. The Wilcoxon signed rank test was used for evaluating the changes over time of the GMFM-88, GMFM-66, and AIMS scores. For correlation analysis, the total scores of GMFM-88, GMFM-66, and AIMS were used. Spearman correlation coefficients were calculated, testing for statistical significance at the α = 0.05 level. Correlation coefficients <0.3 were considered to be weak correlations; those from 0.3–0.7 represent modest correlations, and those >0.7 represent strong correlations. To compare the total scores of GMFM according to the AIMS score, a Mann-Whitney U test was used. The 10th percentile of the AIMS was considered the reference point for delay in motor development, as recommended by the AIMS. The cut-off values for the GMFM-88 and GMFM-66 scores corresponding to the AIMS 10th percentile score for each evaluation period were obtained using a ROC curve with specificity and sensitivity.

III. Results

1. Demographics and birth characteristics

Table 1 provides information about the initial demographic and birth characteristics of the extremely preterm infants in the study. The median GA was 26 weeks (22–27, range 5), and median birth weight was 855 g (490–1980, range 1490). CP was diagnosed in five infants (8.3%), and other major neurologic deficits, including blindness or hearing loss, were diagnosed in two infants (3.3%).

Table 1. Demographic and Birth Characteristic

Parameter	Number (n=60)
Gender (M/F)	25/35
GA (median, min-max)	26weeks (22-27)
IBW (median, min-max)	855gm (490-1980)
Csec (N)	47 (78.3%)
Multiple birth (N)	25 (41.7%)
5min Apgar (median, min-max)	8.00 (1-9)
CP (N)	5 (8.3%)
Major neurological deficits (N)	2 (3.3%)
BPD (N)	31 (51.7%)
PDA (N)	40 (66.7%)
NEC (N)	6 (10.0%)
Sepsis (N)	9 (15.0%)
Cystic PVL (N)	4 (6.7%)

BPD indicates bronchopulmonary dysplagia, requirements for supplemental oxygen for longer than 36weeks of GA; Csec, caesarian section operation; IBW, initial body weight; NEC, necrotizing enterocolitis more than stage 2; PDA, patent ductus arteriosus, the case of operation ; PVL, periventricular leucomalasia.

2. Change of the GMFM and the AIMS scores

The median score of the GMFM-88 was 18.80 (12.0–26.9, range 14.9), and the GMFM-66 was 27.30 (19.7–38.1, range 18.4) at 6 months CA. At 12 months CA, the median score of GMFM-88 was 61.90 (23.4–83.3, range 59.9), the GMFM-66 was 49.62 (31.2–67.0, range 35.8; Table 2). According to the Wilcoxon signed rank test, the GMFM-88, GMFM-66, and AIMS scores all changed significantly over the 6-month follow-up period ($p < 0.001$). The slopes of the two groups were largely similar; however, the slopes were more moderate for the infant with CP. The scores of the infants diagnosed with CP ($n = 5$) were lower than the scores of the infants without CP (n=55) (Table 2). However, there was no statistically significant difference in the scores of GMFM-88, GMFM-66, and AIMS between the infants with or without CP at each evaluation period.

3. Correlation between the GMFM and the AIMS

The total GMFM-88 score was strongly correlated with the AIMS raw score at 6 and 12 months CA ($p < 0.001$). The Spearman correlation coefficient was 0.938 at 6 months CA and 0.725 at 12 months CA. The GMFM-66 score was moderately correlated with the AIMS ($p < 0.001$). The Spearman correlation coefficient was 0.637 at 6 months CA and 0.691 at 12 months CA (Table 3).

4. GMFM according to AIMS percentile score

The 10th percentile of the AIMS was considered the reference point for delayed motor development. There was a significant difference in the GMFM-88 and GMFM-66 total scores between the infants whose AIMS score was above and below the 10th percentile ($p < 0.05$). There were 13 infants whose AIMS score was lower than 10th percentile at 6 months CA, and their mean GMFM-88 and GMFM-66 total scores were 14.80 and 24.70, respectively. These were significantly lower than the GMFM-88 and GMFM-66 scores from infants whose AIMS score were above the 10th percentile (Table 4). Twelve infants had an AIMS score lower than the 10th percentile at 12 months CA. Their mean GMFM-88 score was 52.20, compared to a score

Table 2. Median scores of GMFM-88 and GMFM-66

	6 month			12 month		
	Total (n=60)	Without CP (n=55)	Diagnosed CP (n=5)	Total (n=60)	Without CP (n=55)	Diagnosed CP (n=5)
GMFM-88 (median, min-max)	18.80 (12.0-26.9)	19.50 (12.00-26.90)	16.80 (14.10-22.50)	61.90 (23.4-83.3)	62.05 (31.10-83.30)	39.10 (23.40-56.70)
GMFM-66 (median, min-max)	27.30 (19.7-38.1)	27.30 (19.70-38.10)	25.30 (19.70-28.70)	49.62 (31.2-67.0)	50.09 (40.50-67.04)	38.42 (31.20-49.20)
AIMS (median, min-max)	24 (12-35)	25 (12-35)	17 (14-26)	53 (26-58)	53 (31-38)	38 (26-52)

AIMS indicates Alberta Infant Motor Scale; CP, cerebral palsy; GMFM, Gross Motor Function Measure.

Table 3. Correlation between the GMFM and the AIMS

	AIMS raw score		
	Total Number	Correlation coefficient	p-value
GMFM-88 at 6 month	60	0.938	<0.001
GMFM-88 at 12month	60	0.725	<0.001
GMFM-66 at 6 month	60	0.637	<0.001
GMFM-66 at 12month	60	0.691	<0.001

AIMS indicates Alberta Infant Motor Scale; GMFM, Gross Motor Function Measure.

Table 4. GMFM scores according to AIMS percentile score at 6 month of CA

	6month AIMS score		P-value (Mann-Whitney)
	<10% (N=13)	>10% (N=47)	
GMFM-88	14.80	20.60	
(Median, Min-Max)	(12.00-18.80)	(13.40-26.90)	0.000
(Mean±SD)	15.00±2.337	20.83±3.486	
GMFM-66	24.70	27.30	
(Median, Min-Max)	(19.70-28.70)	(19.70-38.10)	0.023
(Mean±SD)	25.12±2.79	27.60±3.727	

AIMS indicates Alberta Infant Motor Scale; GMFM, Gross Motor Function Measure.

Table 5. GMFM scores according to AIMS percentile score at 12 month of CA

	12month AIMS score		P-value (Mann-Whitney)
	<10% (N=12)	>10% (N=43)	
GMFM-88	52.20	62.30	
(Median, Min-Max)	(23.40-62.80)	(54.50-83.30)	0.000
(Mean±SD)	45.98±10.163	65.43±7.993	
GMFM-66	46.61	50.60	
(Median, Min-Max)	(31.20-52.32)	(46.32-67.04)	0.001
(Mean±SD)	43.62±7.323	52.40±5.014	

AIMS indicates Alberta Infant Motor Scale; GMFM, Gross Motor Function Measure.

of 62.30 for those above 10th percentile. The mean GMFM-66 score was 46.61, and it was also lower than the scores of the infants whose AIMS score were above the 10th percentile (Table 5).

The cut-off values for the GMFM-88 and GMFM-66 scores for detecting delayed gross motor function at each evaluation period were calculated using a ROC curve based on the AIMS 10th percentile score. The calculated cut-off values for GMFM-88 in extremely preterm infants were 17.55 at 6 (sensitivity, 0.809; specificity, 0.846) and 59.05 (sensitivity, 0.737; specificity, 0.800) at 12 months CA. The areas under the ROC curve (AUC) were 0.915 and 0.893, respectively. The cut-off values for GMFM-66 was 25.65 at 6 (sensitivity, 0.702; specificity, 0.615) and 47.10 at 12 months CA (sensitivity, 0.947; specificity, 0.700). The AUC were 0.706 and 0.843, respectively.

Ⅳ. Discussion

This study provides sufficient evidence for the valitidy of the GMFM-88 and GMFM-66 at 6 and 12 months CA in extremely preterm infants born at <28 weeks of GA. There were high correlations between the AIMS scores and the total scores of both GMFM versions. This is the first report demonstrating the validity of the GMFM in extremely preterm infants with varying risk factors, comparing to other standardized gross motor measurement tools for preterm infants. We have suggested cut-off values for the GMFM-88 and GMFM-66 scores at 6 and 12 months CA for extremely preterm infants that can be referenced clinically in evaluating gross motor function.

An overview of the developmental trajectory can provide a better picture of childhood developmental disorders. Although there was not a statistical difference for GMFM-88 or GMFM-66 between infants with or without CP, we observed more gradual slopes in

infants with than without CP. These findings are consistent with previous reports. [3,25] Evaluating infants longitudinally with the same tool is advantageous as it increases the interpretability of interval changes, especially in CP.

With normal motor development, a child should perform all tasks of the GMFM around the age of 5 years. The final achievement level of children with CP depends on the severity of their impairment, and the GMFM scores plateau according to their Gross Motor Function Classification System (GMFCS).[26,27,28] Thus, for children with CP, the upper age limit of the GMFM depends on the child's GMFCS level. However, lower age limits for the GMFM have not been well indicated. The original validation sample of the GMFM-88 only included 21 children aged 5 months–2 years. Therefore, limited information is available on the use of the GMFM during infancy. Some authors reported that scores for both versions of the GMFM do not increase linearly, and they disagree on the applicability of the GMFM for monitoring gross motor development during the early years of life.[20,25,29] Hielkema et al.[25] indicated that the application of the GMFM during infancy is associated with practical problems and may not have a linear pattern in late infancy. Kolobe et al.[29] also concluded that application of the GMFM-88 for children <2 years is hampered by age and developmental characteristics of infancy. Although the present study had only two evaluation points, the scores for both the GMFM-88 and GMFM-66 increased linearly with the AIMS raw scores. The major difference between this and prior studies was the inclusion criteria. As our purpose was to demonstrate the usefulness of the GMFM in extremely preterm infants with very high risk of CP or motor delay, not just for infants with CP, there were fewer infants with CP in the present study than in previous reports. Moreover, the sample size of this study was larger than those of previous studies. Further, all infants were evaluated at the same ages, 6 and 12 months CA.

To avoid the controversy of early use of the GMFM-88, the GMFM-66 was also evaluated. From a Rasch analysis of the GMFM-88, 22 items (13 items in dimension A, five items in dimension B, four items in dimension C) did not fit the unidimensional construct of gross motor function and were eliminated.20) In spite of the limited contribution of dimensions A and B, GMFM-66 still had a moderate correlation with AIMS at 6 and 12 months CA. Hielkema et al.[25] also observed that the GMFM-66 scores improved between 6 and 12 months in 11 of 12 infants; this is in contrast to only minor improvements or deterioration in GMFM-88 scores over the same period. Wei et al.[20] reported that the GMFM-66 had good reliability and validity in assessing the gross motor function of children <3 year old (3–36 months). The observed correlation between GMFM-66 and AIMS in this study is consistent with previous reports. Although the correlation for the GMFM-66 appears weaker than GMFM-88, this suggests a potential advantage using the GMFM-66 in a high-risk infant follow-up program, including CP and motor delays. These results confirm the extended application of the GMFM-88 and GMFM-66 to extremely preterm infants with very high risk for CP or motor delays.

The major limitation of this study was the small number of participants, as this limits the generalization of the results. Other limitations include that this study was restricted to a tertiary care hospital and that it was limited by the retrospective aspect of the design. A last limitation is the relatively short follow-up period, because twenty-four months is a relatively early age for diagnosis of CP. It is possible that a milder type of CP might be diagnosed after a few years in the present cohort. A long-term follow-up and longitudinal assessment should be performed as further studies. Further studies will be required to validate both versions of the GMFM in extremely preterm infants after one year of age compared to other discriminative assessment tools such as the Bayley Scales of Infant Development[30] or the Peabody Developmental Gross Motor Scale[31] for continuous use during childhood. In the case of extremely preterm infants diagnosed with CP, the longitudinal application of the GMFM is not in question, as many previous reports have validated it in children with CP.[20,25,32]

The GMFM-88 and GMFM-66 can be used as valid

measurement tools for evaluating gross motor function in extremely preterm infants at 6 and 12 months CA. Therefore, these scales could be used more widely in early intervention centers or pediatric rehabilitation clinics for longitudinal follow-up of gross motor development in extremely preterm infants.

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