Prospective Longitudinal Study of Gross Motor Function in Children With Cerebral Palsy

Jeanine M. Voorman, MD, Annet J. Dallmeijer, PhD, Dirk L. Knol, PhD, Gustaaf J. Lankhorst, PhD, MD, Jules G. Becher, PhD, MD

ABSTRACT. Voorman JM, Dallmeijer AJ, Knol DL, Lankhorst GJ, Becher JG. Prospective longitudinal study of gross motor function in children with cerebral palsy. Arch Phys Med Rehabil 2007;88:871-6.

Objectives: To describe the course of gross motor function over 2 years in children with cerebral palsy (CP) aged 9 to 15 years, and to investigate its relationship with impairments and age.

Design: Prospective cohort study.

Setting: Rehabilitation department of a university medical center in the Netherlands.

Participants: Seventy boys and 40 girls with CP (mean age \pm standard deviation, $11.2\pm1.7y$).

Interventions: Not applicable.

Main Outcome Measure: The Gross Motor Function Measure (GMFM).

Results: GMFM item scores were stable over the 2 years for the whole group. No difference was found in the course of GMFM item scores between the Gross Motor Function Classification System (GMFCS) levels. We found significant differences in the course of GMFM item scores (corrected for GMFCS) for the different levels of limb distribution, selective motor control, muscle strength, range of motion in the hip and knee, spasticity of the hamstrings, and type of education. There were significantly larger decreases in the more severely affected children. Multivariable analysis showed that a poor selective motor control was the most important determinant of a less favorable course of gross motor function.

Conclusions: Some impairment characteristics may be used to identify children who are at risk for deterioration in gross motor function, and may serve as a guide for interventions.

Key Words: Cerebral palsy; Disabled children; Motor skills; Rehabilitation.

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MOTOR IMPAIRMENTS ARE frequent in cerebral palsy (CP), therefore much of the relevant literature concerns motor functioning. Much attention has been given to the am-

0003-9993/07/8807-11192\$32.00/0 doi:10.1016/j.apmr.2007.04.002 bulatory prognosis and the prognostic factors of ambulation in young children.^{1,2} Even more interesting is whether a child with CP will maintain a certain level of mobility as an adolescent and as an adult. Little is known about motor functioning during puberty and adolescence, but some retrospective studies³⁻⁷ of the motor functioning of adults with CP reported deterioration in mobility and even loss of ambulation in a subgroup of adults with CP. This deterioration results in increased use of adaptive equipment and a greater need for assistance during the activities of daily living.³⁻⁷ These studies reported a relation between the course of motor functioning and the severity of the CP according to the Gross Motor Function Classification System (GMFCS) level, or impairments such as limb distribution and cognitive impairment. Our experience, however, is that some people's mobility deteriorates during puberty and adolescence. Information about changes in motor functioning during adolescence and the factors influencing this prognosis is necessary to establish realistic prognosis and treatment goals that will result in effective use of therapeutic resources and prevent loss of functional abilities. We had 2 main objectives in this study: (1) to describe the course of gross motor function according to level of ability (GMFCS level) over 2 years in children with CP aged 9 to 15 years; and (2) to investigate the relationships between the course of the gross motor function and impairments and age.

METHODS

Participants

Participants were recruited for a 2-year longitudinal study encompassing 3 annual measurements. Rehabilitation centers, special schools for physically and mentally disabled children, and outpatient clinics of departments of rehabilitation medicine in the northwest region of the Netherlands identified 244 children 9, 11, and 13 years of age with CP. These children and their parents received a letter with information about the study and a request that they participate. Of this group, 110 children and their parents returned the informed consent form and participated in the study. Reasons for nonparticipation could be determined in 20 cases: language problems (n=4), moved without a forwarding address (n=2), participation in other research (n=2), and family stress (n=12).

All the regional medical ethics committees approved the study protocol. This research was performed as part of the Pediatric Rehabilitation Research in the Netherlands (PERRIN) program (http://www.perrin.nl), which is a longitudinal study of children with CP.

Data Collection and Outcome Measure

The children and their parents visited the Department of Rehabilitation Medicine at the VU University Medical Center in Amsterdam each year. During the first visit, 2 researchers asked standardized questions about diagnosis, epilepsy, and type of school; classified the children according to the GMFCS; carried out the physical examination; and assessed gross motor

From the Department of Rehabilitation Medicine (Voorman, Dallmeijer, Lankhorst, Becher), EMGO Institute (Voorman, Dallmeijer, Knol, Lankhorst, Becher), and Department of Clinical Epidemiology and Biostatistics (Knol), VU University Medical Center, Amsterdam, The Netherlands.

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Reprint requests to Jeanine M. Voorman, MD, Dept of Rehabilitation Medicine, VU University Medical Center, PO Box 7057, 1007 MB Amsterdam, The Netherlands, e-mail: reva@vumc.nl.

function with the Gross Motor Function Measure (GMFM). All measurements were repeated after 1 and 2 years. Both researchers were certified to administer the GMFM.

The GMFM is a standardized observational instrument that measures gross motor function in children with CP, based on their performance of 88 gross motor tasks upon instruction in a specific test situation. ^{8,9} The GMFM was analyzed with the Gross Motor Ability Estimator (GMAE) computer scoring program to obtain the GMFM-66 score. ^{8,10} The GMAE rescales the child's abilities from an ordinal scale (GMFM-88) to an interval scale (GMFM-66), varying from 0 (poor motor function) to 100 (normal motor function for 5-year-old children).

Determinants

The severity of the CP was classified according to the GMFCS, a 5-level classification system based on functional limitations, the need for assistive devices and, to a lesser extent, quality of movement.^{11,12}

We used the levels of impairments at the first measurement as potential determinants. These levels were limb distribution, selective motor control, muscle strength, limitations in hip and knee extension, spasticity, muscle tone, epilepsy, and type of education. Age was included as a personal characteristic.

Limb distribution was subdivided into 3 categories: hemiplegia (unilateral involvement), diplegia, and tetraplegia (both bilateral involvement). Tetraplegia was defined as the arms being affected as severely or more severely than the legs; diplegia was defined as the legs being more severely affected than the arms. To measure the selective motor control, the children were asked to extend the knee and dorsiflex the ankle of each leg in a short-sitting position without the support of the feet. Possible scores were: 0 (no selective, only synergistic movement), 1 (diminished selective movement [the first range of movement selective and later on, during the movement, no selective movement]), and 2 (full selective movement during extension of the knee and dorsiflexion of the ankle). The scores for the 2 sides together produced a total score varying from 0 to 8. The total scores were then subdivided into 3 categories: poor selective motor control (total scores 0, 1, or 2); moderate selective motor control (total scores 3, 4, or 5); and good selective motor control (total scores 6, 7, or 8).

To define muscle strength, the children were asked to stretch out from squat position 8 times (support for balance was allowed). They were subdivided into 3 categories: good strength if they could squat 8 or more times, moderate strength if they could squat fewer than 8 times or performed a part of the motion 8 times, and poor strength if they were not able to squat at all.

The range of motion (ROM) of hip and knee extension was measured, both in a supine position. ¹³ We performed the Thomas test¹⁴ to detect the limitations in hip extension. To indicate the degree of limitations in ROM, the ROM scores were transformed according to the Spinal Alignment and Range of Motion Measure (SAROMM), to discriminate between no (1), mild (2), moderate (3), and severe (4) limitations in ROM (see appendix 1). ¹⁵ To indicate the total extent of the limitations in extension in hips and knees we calculated an overall score as the mean of the SAROMM scores of the knee and hip of the right and left legs.

Spasticity was measured as an increase in muscle tone resulting in a catch during fast velocity stretching of the muscles, using standardized measurement procedures. ¹⁶ Spasticity in the hamstrings, hip adductors, and the gastrocnemius muscle was measured in a supine position, and spasticity in the rectus femoris muscle was measured in a prone position. The scores

were as follows: 0 if no spasticity was found in a muscle group, 1 if spasticity was found on one side, and 2 if the muscle group was found to be spastic on both sides. We calculated an overall score to indicate the total extent of the spasticity as the mean of the spasticity in the 4 muscle groups.

The muscle tone was measured as resistance in slow stretching during the ROM measurements and was defined as normal or abnormal if more than 50% of the muscles were hypotonic or hypertonic.

Children with more than 1 seizure during the previous 2 years were defined as having repeated seizures.

Type of education was based on the type of school: children with a "regular" education were those in a regular school or in a school providing education for physically disabled children, whereas children with "special" education were those enrolled in special schools for children with cognitive impairment (with or without physical disabilities), or in special day-care centers for severely handicapped children.

Finally, the children were subdivided into 3 age groups: children who were 9, 11, or 13 years of age at the first measurement.

Statistical Analyses

We used random coefficient analyses, also known as multilevel analysis (MlwiN^{17,a}), to analyze the changes in the GMFM over time and its determinants. This analysis method considers the dependency of repeated measures within the same person by allowing the regression coefficients to differ between subjects. In addition, the number of observations per person may vary (ie, subjects with missing values can be analyzed). The data were defined as follows: level 2 as patient and level 1 as measurement occasion. The GMFCS, limb distribution, selective motor control, muscle strength, ROM, spasticity, and age were analyzed as categorical variables using dummy variables. Type of education, epilepsy, and muscle tone were analyzed as dichotomous variables. Time was expressed as the measurement occasion in years.

Time, the GMFCS, and the interaction term GFMCS by time were added to the model to analyze the course of gross motor function, as described above. To analyze the relation between the course of gross motor function with impairments and age, each of the determinants and the interaction terms with time were entered into the model separately, corrected for GMFCS level. Subsequently, we made a multivariable model using a forward stepwise procedure, beginning with the most significant determinant. First, each determinant was added to the model as a single factor and then removed if not significant (P>.05). After this, the interaction action terms were added to the model one by one and then also excluded if not significant (P>.05). We used the chi-square test to determine whether determinants were significant (P<.05).

To determine the required sample size, we did a power analyses, using an α of .05, power of 0.8, smallest meaningful difference over a year of 2 points change on the GMFM, and a variance of 54. For the dependency of the repeated measures we assumed an intraclass correlation coefficient of .90. This resulted in a sample size of at least 11 subjects for each GMFCS group.

RESULTS

Table 1 shows the children's baseline characteristics. Of the 110 participating children, 6 were lost to follow-up after 1 year and 5 after 2 years (GMFCS I: n=3; GMFCS II: n=3; GMFCS III: n=3; GMFCS IV: n=2; 8 boys, 3 girls; mean age ± standard deviation, 10.28±1.34y). In addition, 1 child who missed the second measurement participated in the third.

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