Early Human Development 95 (2016) 9–13

Contents lists available at ScienceDirect

Early Human Development

journal homepage: www.elsevier.com/locate/earlhumdev

Specific characteristics of abnormal general movements are associated with functional outcome at school age

Elisa G. Hamer [a,c], Arend F. Bos [b], Mijna Hadders-Algra [a,*]

[a] Department of Pediatrics, Division of Developmental Neurology, University of Groningen, University Medical Center, Groningen, The Netherlands
[b] Department of Pediatrics, Division of Neonatology, University of Groningen, University Medical Center, Groningen, The Netherlands
[c] Department of Neurology, Radboud University Medical Center, Nijmegen, The Netherlands

ARTICLE INFO

Article history:
Received 12 August 2015
Received in revised form 13 January 2016
Accepted 19 January 2016

Keywords:
General movements
High risk infants
Cerebral palsy
Follow-up
School age
Neurodevelopment

ABSTRACT

Background: Assessing the quality of general movements (GMs) is a non-invasive tool to identify at early age infants at risk for developmental disorders.
Aim: To investigate whether specific characteristics of definitely abnormal GMs are associated with developmental outcome at school age.
Study design: Observational cohort study (long-term follow-up).
Subjects: Parents of 40 children (median age 8.3 years, 20 girls) participated in this follow-up study. In infancy (median corrected age 10 weeks), the children (median gestational age 30.3 weeks; birth weight 1243 g) had shown definitely abnormal GMs according to Hadders-Algra (2004). Information on specific GM characteristics such as the presence of fidgety movements, degree of complexity and variation, and stiff movements, was available (see Hamer et al. 2011).
Outcome measures: A standardised parental interview (presence of CP, attendance of school for special education, Vineland Adaptive Behavior Scale to determine functional performance) and questionnaires (Developmental Coordination Disorder Questionnaire [DCD-Q] to evaluate mobility and Child Behavior Checklist to assess behaviour) were used as outcome measures.
Results: Six children had cerebral palsy (CP), ten children attended a school for special education, and eight children had behavioural problems. Both the absence of fidgety movements and the presence of stiff movements were associated with CP ($p = 0.001$; $p = 0.003$, respectively). Stiff movements were also related to the need of special education ($p = 0.009$). A lack of movement complexity and variation was associated with behavioural problems ($p = 0.007$). None of the GM characteristics were related to DCD-Q scores.
Conclusions: The evaluation of fidgety movements and movement stiffness may increase the predictive power of definitely abnormal GMs for motor outcome — in particular CP. This study endorses the notion that the quality of GMs reflects the integrity of the infant's brain, assisting prediction of long-term outcome.

1. Introduction

In early infancy, the assessment of general movements (GMs) is the best clinical predictor for neuromotor development in high risk infants [1,2]. GMs are endogenously generated spontaneous movements and their quality reflects the integrity of the infant's brain [3]. GMs are present from early foetal life until goal-directed motor behaviour arises, i.e., typically around 4 months corrected age. Normal GMs are complex movements in which all body parts participate; they are characterised by variation [4]. In abnormal GMs, complexity and variation are reduced. The presence of definitely abnormal GMs is associated with perinatal brain lesions and developmental disorders, including cerebral palsy (CP) [1,5].

Recently, we were able to demonstrate that specific movement characteristics might enhance prediction of definitely abnormal GMs (classified according to Hadders-Algra, 2004 [4]) around 3 months of age, i.e., so-called fidgety-GM age [6]. Both the absence of fidgety movements and the presence of predominantly stiff movements were associated with a higher chance of CP at 18 months. None of the other movement characteristics investigated were associated with functional motor performance or cognitive outcome at that age.

It is known that associations between early neurological signs and developmental outcome may change with age, as the child's brain

Abbreviations: ATNR, Asymmetric tonic neck reflex; CBCL, Child Behavior Checklist; COPCA, Coping with and caring for infants with special needs; CP, Cerebral palsy; DCD-Q, Developmental Coordination Disorder Questionnaire; DSM, Diagnostic and Statistical Manual of Mental Disorders; GMs, General movements; VABS, Vineland Adaptive Behavior Scale.

* Corresponding author at: University Medical Center Groningen, Department of Developmental Neurology, Hanzeplein 1, 9713 GZ Groningen, The Netherlands. Tel.: +31 50 3614247; fax: +31 50 3619158.
E-mail address: m.hadders-algra@umcg.nl (M. Hadders-Algra).

http://dx.doi.org/10.1016/j.earlhumdev.2016.01.019
0378-3782/

experiences significant changes during development [7,8]. Also, the age of 18 months corrected age, used in Hamer et al. 2011 [6], is relatively early for the diagnosis of CP. For instance, the Surveillance of Cerebral Palsy Europe only includes children older than 5 years of age [9]. Moreover, 18 months corrected age is early in terms of outcome in other domains, e.g., functional and behavioural outcomes.

The primary aim of the present study is therefore to investigate whether the specific characteristics of definitely abnormal GMs that were related to motor outcome at 18 months, i.e., fidgety movements and predominantly stiff movements (further denoted as stiff movements), are related to developmental outcome at school age. In addition, we studied whether the other movement characteristics studied in Hamer et al. [6], such as the degree of complexity and variation, were associated with outcome at school age.

2. Methods

This study is part of the VIP-project (Dutch; Vroegtijdig Interventie Project), a randomised controlled trial studying the effects of the early intervention program COPCA (Coping with and Caring for infants with special needs — a family centred program [10]) in comparison to traditional infant physiotherapy. The developmental outcome in the two intervention groups was similar [11]. Inclusion in the VIP-project was based on the presence of definitely abnormal GMs [4]. The general movement assessment includes at least 5 min of video recording of spontaneous motor behaviour in an awake, non-crying state. As previously reported, MHA and AFB reanalysed the video recordings of the VIP infants ($n = 46$, median age at GM assessment 10.3 [range 9–13] weeks corrected age) with respect to the following characteristics: presence of fidgety movements, degree of complexity and variation, presence of cramped-synchronised GMs, stiff or jerky movements, and spontaneous occurrence of the asymmetric tonic neck reflex (ATNR) pattern (Table 1) [6]. In addition, we computed a movement abnormality score (MAS; range 0–2) based on the absence of fidgety movements (1 point) and the presence of stiff movements (1 point).

2.1. Follow-up assessment at school age

We sent an invitation letter for the present follow-up study to the parents whose children participated in the follow-up assessment at 18 months ($n = 44$). For the follow-up at school age, we used a structured parental telephone interview (Vineland Adaptive Behavior Scale [VABS], medical history, educational achievement) and parental questionnaires (Developmental Coordination Disorder Questionnaire [DCD-Q] and Child Behavior Check List [CBCL]). During the interview the parents specified the child's educational achievement, including the need of special education. Notwithstanding the fact that the Dutch government is in favour of inclusive education, children with special needs often attend schools for special education. Both the parents and the interviewer (EGH) were unaware of the specific general movement characteristics of the child.

Using the VABS we assessed the child's functional status in four domains: communication, daily living skills, socialisation and motor skills [12]. The four domains can each be subdivided in 2–4 subdomains, e.g., expressive communication, play and leisure time, and fine motor skills. We calculated the scores for each domain ($n = 4$) and subdomain ($n = 11$). The VABS has proven to be reliable and valid in both typically developing children and in children with developmental disorders, such as CP [13,14]. The domain communication, daily living skills and socialisation can be applied until 18 years of age. The motor skill domain is designed for children up to 6 years of age and for older children with motor impairments.

We used the DCD-Q to assess mobility [15]. This short questionnaire covers 17 items on control during movement, fine motor skills/handwriting, gross motor skills, and general coordination. A higher total score indicates worse performance. The DCD-Q is reliable and valid, as is the Dutch translation [16].

The CBCL is a widely used parental questionnaire containing 113 items concerning the child's behaviour [17]. The scores were dichotomised as normal or borderline vs. in the clinical range (i.e., above the 97th percentile) on the following six Diagnostic and Statistical Manual of Mental Disorders (DSM) oriented scales: affective, anxiety, somatic, attention deficit/hyperactivity, oppositional defiant, and conduct problems. The reliability and validity of the Dutch version of the CBCL are good [17,18]. The Medical Ethics Committee of the University Medical Center Groningen approved the study (trial number NL39954.042.12).

2.2. Data analysis

Statistical analysis was performed using SPSS version 20. We used non-parametric tests since the data did not show a normal distribution. Regression analysis was used to control for type of intervention. To adjust for multiple comparisons, differences with a p-value ≤ 0.01 were considered to be statistically significant. Psychometric properties and confidence intervals (CIs) were calculated with the use of MedCalc statistical software (version 11.6, Ostend, Belgium).

3. Results

We obtained follow-up data from 40 children (91%, median age; 8 years + 4 months; Table 2). The participating children did not differ from the non-participating children in baseline characteristics (sex, gestational age, birth weight, educational level of the parents, maternal age at birth, type of intervention or CP at 18 months CA — data not shown).

3.1. GM characteristics: Motor and functional outcome

Six parents reported that their child had CP. Both the absence of fidgety movements and the presence of stiff movements were associated with CP (Table 3, Fisher's exact test; $p = 0.001$ and $p = 0.003$, respectively). The absence of fidgety movements and the presence of stiff

Table 1
Specific GM characteristics.

Fidgety movements	Fidgety movements are circular movements of small amplitude and moderate speed and variable acceleration of neck, trunk and limbs in all directions.
Complexity and variation of the movements	Complex movements are movements during which the infant actively produces frequent changes in direction of the participating body parts. The GM variation represents the temporal variation of the movements. This means that across time, the infant produces continuously new movement patterns.
Cramped-synchronised movements	Cramped-synchronised movements appear rigid and lack normal smooth and fluent character; all limb and trunk muscles contract and relax almost simultaneously.
ATNR pattern	The asymmetric tonic neck reflex (ATNR) consists of the extension of the arm and leg on the side to which the head is turned, with flexion of the contralateral limbs.
Stiff or jerky movement quality	GM fluency indicates the presence of smooth, supple and graceful movements. Stiff movements and jerky movements are a type of non-fluent motor behaviour. Non-fluent movements can be predominantly jerky, stiff or consist of a mix of jerky and stiff movements.

For details on the scoring of the specific GM-characteristics see Hamer et al. [6].