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General movement assessment: Predicting cerebral palsy in clinical practise

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Abstract

Objective: The general movement assessment (GMA) method is used to predict cerebral palsy (CP) in infants with high risk of developing neurological dysfunctions. Most of the work on GMA has been performed from the same group of researchers. The aim of this study was to demonstrate to what extent GMA predicted CP in our hands.
Method: A prospective study was performed using the Prechtl classification system for GMA in the fidgety period to predict later cerebral palsy. The study population consisted of 74 term and preterm infants at low and high risk of developing neurological dysfunction. The absence or presence of CP was reported at 23 months median-corrected age by the child's physician and the parents.
Results: The GMA identified all 10 infants that later were classified as having CP. GMA also identified all the infants that did not develop CP except for one infant with abnormal GMA and no CP. Three infants had uncertain CP status at follow-up. The sensitivity of GMA with regard to later CP was 100% with 95% CI (0.73, 1.00) and the specificity was 98% with 95% CI (0.91, 0.99) when the three uncertain cases were excluded.
Conclusion: Our study indicates that the GMA used in a clinical setting strongly predicts the development of CP. The work supports the results of previous studies and contributes to the

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validation of GMA. The qualitative nature of this method may be a problem for inexperienced observers. Larger clinical studies are needed.

1. Introduction

Despite technical advances and improvements in obstetric and neonatal care over the last two decades, the prevalence of cerebral palsy (CP) remains constant [1]. Survival among extremely low birth weight infants with a high risk of CP has increased, whereas improvements in perinatal care may have led to a small, but significant decrease in CP among term infants [2]. The diagnosis of CP is usually not established until late in the first year of life [3], and mild cases may not be diagnosed until the age of four or even later [4]. Early prediction of CP is considered important in directing appropriate intervention programs and in identifying those children in need of close surveillance [5,6].

Clinical evaluation of newborn infants in order to predict later neurological disabilities is difficult. Dubowitz et al. [7], Prechtl [8] and Amiel-Tison and Grenier [9] have described some well-known neonatal neurological assessment tests. All of these tests are based on the assessment of passive and active muscle tone and a number of elicited reflexes and reactions. The ability of each test to predict neurological outcome in preterm and term infants varies in different studies [8,10–12]. In addition to clinical examination, imaging of the newborn brain with cerebral ultrasound (CUL) and magnetic resonance imaging (MRI) has improved the prediction of neurological outcome in high-risk infants [13,14].

Prechtl and co-workers have studied a special type of spontaneous movements in newborns and small infants, the so-called general movements (GM). Unlike reflexes, spontaneous movements are patterns of movements that are not initiated by any obvious external stimuli. Observation of the infant's GM and especially the so-called fidgety movements (FMs) has shown promising scientific results with regard to prediction of later neurological impairment [10,15]. FMs may be seen at 6 to 20 weeks post-term and are normally present at 10–15 weeks post-term [5]. Lack of normal fidgety movements has been shown to predict neurological outcome at 2 years more precisely than standard neurological examination both in high-risk preterm infants and in term infants with hypoxic–ischaemic encephalopathy [16–18]. Inter-observer reliability varies from 78% to 93% [5,19–21].

Although promising, many questions remain regarding the implementation of GMA in standard clinical practise. Most of the studies on GMA have come from a few groups of researchers, and the generalizability of the GMA as a clinical tool has been questioned [3]. The methodology has a qualitative approach, and classifications are made based on subjective judgements. Professional training, background knowledge about the child's medical history and frequency of observations may influence the evaluation of the GMs.

For several years GMA has been used to evaluate infants at risk for neurological impairment at St. Olavs Hospital, Trondheim University Hospital. The method is used in addition to standard neurological examination and other available techniques including cerebral ultrasound and MRI. The aim of this study was to evaluate, in this clinical setting, to which extent GMA performed during the fidgety period, predicted CP.

2. Subjects and methods

2.1. Subjects

The majority of infants enrolled were from St. Olavs Hospital. High-risk infants (term and preterm) and low-risk preterm infants were included from the neonatal intensive care unit, whereas healthy term infants were included from the maternity ward. In addition, nine high-risk infants were included from four other hospitals in Norway. High-risk infants were included based on the medical history and cerebral ultrasound results. Children were classified into the high-risk group if they had one or more well-known perinatal risk factors for neurological impairment (Table 1). Infants with congenital syndromes and malformations that could interfere with their spontaneous movements were excluded from the study. Only infants with GMA performed at 10–18 weeks post-term were included.

Neuroimaging results from the neonatal period were collected on all high-risk infants. All units involved in the study did sequential US scans on extremely low birthweight infants in the neonatal period. The timing and frequency of US examinations was in accordance with the different unit's own protocols. Magnetic resonance imaging and CT were available to all units involved and were done at the attending physician's discretion.

Table 1 Criteria for high-risk classification of pre-term and term babies

Criteria	Preterm[a] (*n*)	Term[a] (*n*)
Perinatal stroke[b]		3
Perinatal asphyxia[c]		5
Intra-/peri-ventricular hemorrhage (IVH/PVH), grade III or IV	7	
Severe hypoglycemia and *E. coli* sepsis	1	
BW <1000 g and/or GA <28 weeks	14	
Bronchopulmonary dysplasia with suppl. O_2 at discharge	2	

[a] Some of the infants had more than one risk factor.
[b] Perinatal stroke: Two patients with arterial stroke and one patient with a haemorrhagic infarct after open-heart surgery.
[c] Perinatal asphyxia: All five needed assisted ventilation after resuscitation for from 10 min to several days. Three had Apgar scores ≤3 at 5 min. Three developed neonatal seizures with HIE grades II–III and one of these had MRI findings consistent with hypoxia/ischemia in the neonatal period. Two had non-specified signs of perinatal stress and HIE grade I.

2.2. Observation of general movements

The GMA using video recordings were performed 10—18 weeks post-term in order to study the absence or presence of normal fidgety movements. Recordings were performed according to the standard method for GM observation [20], at least 30 min after feeding and lasted for several minutes during periods of active wakefulness. The infant was partially dressed (body vest and nappy), lying supine. The temperature in the room was comfortable (24—28 °C) and the infant had enough space to move spontaneously. The recordings were repeated several times (range 1—5) to ensure that the quality of movements (normal or abnormal) could be accurately judged.

Fidgety movements were defined according to Prechtl as circular movements of small amplitude, moderate speed, and variable acceleration of neck, trunk and limbs in all directions [5]. Normal fidgety movements are characterized as a continuous stream of tiny and elegant movements [10] and were classified as normal when they were present (F+, isolated events, or F++, continuous). Fidgety movements were classified as abnormal if they were absent (F−) or abnormal in nature; looked like normal fidgety movements but their amplitude, speed and jerkiness were moderately or greatly exaggerated (Fa) [5]. The video recordings were edited to include representative samples of movements lasting from 30 s to several minutes for each infant.

All recordings were performed and classified by the same physiotherapist (LA), who also had knowledge of the medical history of the infants. The physiotherapist had participated in GMA basic and advanced training courses and had 4 years of clinical experience in using GMA. He was certified by the General Movement Trust (GMT) performing the Prechtl methodology. In order to test inter-observer reliability, a GMA-trained physiotherapist (GKØ) from a different hospital, who was unaware of both the medical history of the infants and the initial GMA classification, performed a second GMA classification of the same recordings. This observer had also participated in basic and advanced training courses and had certification from the GMT and several years of clinical experience using the Prechtl methodology.

2.3. Neurological outcome at 2 years of age

All infants in the high-risk group enrolled from St. Olavs Hospital (16 of the 25 high-risk infants included in the study) had follow-up at the hospital's outpatient program for young children at risk of neurological adverse outcome. A multidisciplinary team assesses the child at 3, 9, 15 and 24 months corrected age and at 5 years of age before starting school. The team includes a consultant in neonatology, a child physiotherapist, an occupational therapist, a specialist in neuropsychology and a special education therapist. The same consultant in neonatology (RS) did clinical neurological examination of all children, and motor and mental skills were assessed using validated tests (AIMS test at 9 and 15 months and Bayley score for motor and mental function at 24 months).

Of the 9 children who had follow-up at other hospitals, 5 were followed at institutions with similar structured, multidisciplinary follow-up programs. Four children had follow-up at a hospital where no structured follow-up program had been implemented, but where the same, experienced paediatrician and a child physiotherapist were responsible for follow-up and other subspecialties were involved on clinical indication. For all the low risk infants, information regarding CP status was obtained from the public health nurse and/or family physician, as none of these children had routine contact with a paediatrician.

In order to ascertain that all children with a potential motor problem were identified, all parents were asked to fill out a questionnaire about whether their child had CP or not. Based on all this information, neurological outcome for each child was classified into three groups: cerebral palsy, not cerebral palsy or uncertain.

2.4. Ethics

All infants included in the present study had participated in a previous study were parents had approved video recordings of spontaneous movements of their child. Before the present study, all parents received a letter asking their informed consent for their child to participate in this follow-up study. When they approved to participate in the study, they also allowed the investigators access to the medical records of their children and to contact their local health professionals. Parents who did not respond were reminded first by telephone and finally by a letter. Physicians and public health nurses were reminded by a telephone call. The study was approved by the Regional Committee for Medical Research Ethics and Norwegian Social Science Data Services.

2.5. Statistical analysis

Outcome data were compared with data collected from the GMA analysis. Statistics were carried out using the program StatXact-5 (5.0.3). A confidence interval of 95% for sensitivity and specificity were calculated.

3. Results

3.1. Study population

Of the 83 letters sent to the parents, 79 were returned. Four families did not give their consent to contact their family physician and/or the public health nurse. The remaining 75 parents approved to participate in the follow-up study. Of the 75 letters sent to the family physicians and the public health nurses, 74 answers were returned. The final study population consisted of these 74 children (33 boys and 41 girls). Forty-two (57%) infants were born preterm (Table 2). In the preterm group, the median gestational age was 30.5 weeks (range 24—36 weeks) and median birth weight was 1367 g (range 540 to 3800 g). None of the infants in the study group were born after 42 weeks. Among preterm infants 40% were classified as high-risk, whereas 25% were classified as high-risk among the term infants (Table 2).

Of the 25 high-risk infants, 7 had major abnormalities on US defined as IVH grade III—IV with or without PVL. One of

Table 2 Term and preterm infants classified in high-risk and low-risk groups ($n=74$)

Gestational age	High-risk	Low-risk	Total
Term (≥37 weeks)	8	24	32
Preterm (<37 weeks)	17	25	42
Total	25	49	74

these also had congenital hydrocephalus and later developed ventriculitis. One infant had MRI changes consistent with hypoxic/ischemic encephalopathy, two infants had arterial infarcts diagnosed on MRI or CT and one infant had a haemorrhagic infarct diagnosed on MRI. Three infants had minor abnormalities on cerebral US defined as IVH grade I—II. Eleven of the infants in the high-risk group were classified with normal US and/or MRI in the neonatal period.

3.2. Quality of general movements

One hundred and thirty-five GM assessments were performed in the fidgety period between 10 and 18 weeks post-term in the 74 infants (range 1 to 5 assessments per infant). Sixteen of seventeen children with assessments between 16 and 18 weeks post-term also had assessments earlier in the fidgety period. The one child with only one late assessment (at 18 weeks post-term) had abnormal GMA and had CP on follow-up. Observer 1 (LA) performed the initial classification on which the calculations of sensitivity and specificity were based. All FMs judged as abnormal by this observer were identified as absent (F—) and none as abnormal in nature (Fa). In the high-risk group, 12 of the 25 infants were classified as F—, whereas only 1 infant of the 49 infants in the low-risk group was classified as F—.

3.3. Neurological outcome

At follow-up, 10 children had CP, 61 had no CP and three had an uncertain CP status (Table 3). Classifications by health professionals were based on medical information from the child's last consultation at a corrected median age of 23 months (range 9—31 months). Median age of children at follow-up based on the parents report was 26 months corrected age (range 9—34 months). The ten children with CP were classified with full consistence between health professionals and the parent's report. Two infants with a follow-up of only 9 months both had definite CP. The shortest follow-up, except from these two, was 13 months. Sixty-one children were classified with no CP both by professional health workers and by the parents. Two of three children with uncertain CP status were classified as uncertain by both health professionals and the parents, whereas one was classified as uncertain by the parents and as not having CP by the paediatrician. For these three children, a telephone call to the paediatrician 6 months later (at 25, 32 and 32 months follow-up, respectively) revealed a normal outcome for one, an uncertain outcome for one and one with CP.

3.4. Prediction of neurological outcome

Of the 61 infants with normal FMs classified by observer 1, 60 did not develop CP (Table 3). One child with normal GMA was classified as having uncertain CP status at follow-up at 25 months corrected age, whereas 6 months later his CP status was changed to no CP according to the attending paediatrician. Among the 13 infants with abnormal GMA, 10 were diagnosed as having CP at follow-up (Table 3). Of the three remaining infants with abnormal GMA, one had no CP and two had an uncertain CP status (6 months later these two were classified as one with CP, one still uncertain). In the high-risk group, 40% of the infants developed CP and none in the low-risk group (Table 4). The 10 infants with CP are described in details in Table 5.

3.5. Inter-observer comparative classification

An additional GMA classification was performed by a physiotherapist from another hospital (observer 2) in 73 of the 74 children. She was unaware of the initial GMA classification and the medical history of the infants. The classification was identical between observer 1 and 2 in 64 infants (87.7%) and different in 9 infants. Four infants were classified as having fidgety movements by observer 1 and having no fidgety movements by observer 2. Three infants were classified with no fidgety movements by observer 1 and as having fidgety movements by observer 2. Two infants classified with FMs that looked abnormal in nature (Fa) by observer 2 were both classified as normal FMs by observer 1. These two had no CP on follow-up. Three out of the remaining seven infants with different GMA classification had uncertain neurological outcome at follow-up. The inter-scorer agreement (Cohens Kappa) resulted in κ value of 0.61 with 95% CI (0.37, 0.84) for the 73 children.

3.6. Sensitivity and specificity

By leaving out the three children with uncertain CP status, the sensitivity and specificity were calculated for 71 out of 74 children with a definite outcome for observer 1 and for 70 out of 73 children for observer 2. For observer 1 the sensitivity was estimated to 1.0 with 95% CI (0.73, 1.00) and specificity to 0.98 with 95% CI (0.91, 0.99) and for observer 2 the sensitivity was estimated to 1.0 with

Table 3 Classification of neurological outcome in relation to fidgety movements ($n=74$)

Quality of fidgety movements	CP	No CP	Uncertain	Total
Abnormal	10	1	2	13
Normal	0	60	1	61

Table 4 Classification of neurological outcome in relation to risk group ($n=74$)

Risk classification	CP	No CP	Uncertain
High-risk	10	12	3
Low-risk	0	49	0
Total	10	61	3

Table 5 Children with CP at follow-up ($n = 10$)

Case	GA (week)	FV (g)	Risk factors	CP type
1	40	3570	Arterial infarct	Right hemiplegia
2	29	920	IVH grade IV, congenital hydrocephalus, ventriculitis	Right hemiplegia
3	41	3456	Perinatal asphyxia (Apgar 1-4-7, HIE grade 2—3, neonatal seizures, assisted ventilation)	Quadriplegia
4	27	565	IVH grade IV	Right hemiplegia
5	41	3790	Perinatal asphyxia (Apgar 0-0-2, HIE grade II, neonatal seizures, assisted ventilation)	Quadriplegia
6	40	3490	Perinatal asphyxia (Apgar 2-3-6, HIE grade II—III, neonatal seizures, assisted ventilation)	Quadriplegia
7	40	3580	Haemorrhagic infarct after open heart surgery	Left hemiplegia
8	24	717	IVH grade IV	Right hemiplegia
9	24	695	IVH grade IV	CP, unspecified
10	34	1740	Severe hypoglycemia and *E. coli* sepsis	Quadriplegia

95% CI (0.72, 1.0) and specificity to 0.92 with 95% CI (0.82, 0.96).

4. Discussion

The analysis of general movements has been described as a sensitive method to predict later neurodevelopmental disorders in infants. Although the method has been in use for more than 10 years, there are still few reports on its application from outside the scientific groups where it was first described. In this study, we wanted to see if GMA used in a clinical setting, could predict later CP. The GMA classification was not compared to other tests for neurodevelopmental prediction, and the physiotherapist performing the GMA classification was aware of the medical history of the infants.

General movement assessment performed during the fidgety period identified all infants that later developed CP. Furthermore, normal FMs correctly identified infants that did not develop CP apart from one child. These results support that GMA is a good method to identify those at risk for developing CP [5,16,22], and that normal FMs in high-risk infants can be used to predict a low risk of developing CP.

In a study, by Prechtl et al. [5], a mixture of high- and low-risk infants was included, similar to the approach in the present study. In that study, fidgety movement assessment predicted CP with a sensitivity of 95% and a specificity of 96%. Regardless of the different etiological factors predisposing for CP, it appears that abnormal FMs is a common phenomenon for infants that later develop CP.

In the present study neurological outcome was assessed at a median corrected age of 23 months. This corresponds well with other studies [16,17]. An experienced paediatrician followed all high-risk infants, and majority of high-risk infants were enrolled in a structured follow-up program for young children at risk of neurodevelopmental adverse outcome. However, the range of age at follow-up was wide (13—31 months median corrected age when two infants with CP classified at 9 months were not taken into account). Milder forms of CP may present later in childhood, leading to an underestimation of CP in this study. Although GMA appears to be a good method to predict CP which presents at an early age, it is still an unanswered question if GMA is equally good in predicting outcome in those with a milder form of CP.

Follow-up in the low-risk group was based on information by health professionals who are not necessarily trained to detect subtle neurological symptoms in very young children. However, the prevalence of CP in the general population is as low as 1.50—3.00 per 1000 live births [4]. The likelihood of any of the 49 children in this group having CP, despite a normal development as judged by a public health nurse/family physician and parents at 26 months of age, is therefore very low.

Three infants had an inconclusive clinical outcome. All these three also had non-concordant GMA classifications by the two observers. Two of the nine infants with non-concordant GMA classifications were classified with abnormal fidgety movements (Fa) by observer 2 and with normal FMs by observer 1. The long-term outcome of infants with fidgety movements that look abnormal in nature (Fa) is less clear and may turn out to be cerebral palsy, developmental retardation or minor neurological dysfunctions [5]. In a 3-year follow-up study of 16 infants with Fa classification, three turned out normal, seven showed evidence of developmental retardation or minor neurological signs and six developed CP [5]. The present study was not designed to detect neurological dysfunctions other than CP. It is, therefore, not possible to know if children who were classified differently by the two observers will develop other "soft" neurological signs at an older age.

Knowing the medical history of the infants may have biased the judgements of the physiotherapist in our study (observer 1). An inter-observer concordance of 87.7% as reported in this study is in accordance with that reported by others [20]. Agreement between observers in almost 90% of cases makes it unlikely that the initial classification was significantly biased.

The most frequent abnormality on neonatal cerebral ultrasound in high-risk infants in this study was the presence of IVH grade III—IV, whereas none were diagnosed with cystic PVL in the absence of IVH. This is most likely due to the way infants were recruited. In order to validate the GMA method with regard to prediction of CP, high-risk infants were recruited if they were considered at high risk of motor impairment based on the presence of major US abnormalities or MRI findings or the clinical history. Intraventricular haemorrhage can be easily detected on early US scans, whereas the detection of cystic PVL may depend on serial US

scans beyond the first 28 days of life [14]. One child with IVH grade I and one child with normal neonatal US scan had MRI findings at 4 years of age consistent with PVL. This suggests that a diagnosis of PVL may have been missed and that infants with PVL were not included in the present study unless they had other major risk factors that made them eligible.

The GMA is non-invasive, cheap and independent of advanced technical equipment. Performing the GMA does not seem to put the child in a stressed situation. The most experienced GM assessors claim that the GMA is easy to learn and easy to perform [5,10]. In most studies published about GMA, the observers have been very experienced. The physiotherapists in this study were also experienced, and it is therefore still an unanswered question how much experience the observer needs to make a valid clinical assessment. The whole procedure, including video recording, editing and classification, takes approximately one hour per assessment. In addition, repeated assessments are often necessary to perform an optimal GMA. An informal telephone interview to some of the larger neonatal intensive care units in Scandinavia, revealed that although the neonatologists and physiotherapists had a fair theoretical knowledge of GMA, very few used it in clinical practise. This indicates that more scientific documentation and validation in clinical trials is needed.

5. Conclusion

Although small, this study indicates that GMA, used in a clinical setting in a high-risk population, can be a useful tool to predict later CP. The study supports the results of previous studies and contributes to the validation of GMA. More studies in larger populations are needed to verify the results, especially in predicting mild CP.

The qualitative nature of this method may be a problem for clinicians working alone, implying a risk of drifting away from the standards of the methodology. An aim for future studies is therefore the development of more objective classification criteria and a standardised way of analysing spontaneous movements.

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