

QUALITY OF GENERAL MOVEMENTS AS A MEANS TO EVALUATE THE INTEGRITY OF THE YOUNG NERVOUS SYSTEM

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Detection of children with a developmental disorder, such as cerebral palsy, at early age is notoriously difficult. Recently a new form of neuromotor assessment of young infants has been developed, based on the assessment of the quality of General Movements (GMs). GMs are movements of the fetus and young infant in which all parts of the body participate. The hallmark of typical GMs is movement complexity and variation; in abnormal GMs movement complexity and variation is reduced or absent. Recently it has been hypothesized that GM complexity and variation are brought about by the transiently present cortical subplate and that abnormal GMs are the result of damage or dysfunction of the subplate and its efferent motor connections in the periventricular white matter. Multiple studies indicated that abnormal GMs may predict developmental outcome. Prediction on the basis of longitudinal series of GM-assessments is best. Second best is prediction on the basis of an assessment at "fidgety" GM age, i. e. at 2-4 months post-term. Definitely abnormal GMs at "fidgety" age are related to cerebral palsy, mildly abnormal GMs to minor neurological dysfunction at school-age. This implies that definitely abnormal GMs at fidgety age are an indication for early physiotherapeutic intervention.

Descriptors: GENERAL MOVEMENTS, INFANCY, PREDICTION, EARLY DETECTION, CEREBRAL PALSY

Introduction

Prediction of developmental outcome in early infancy is difficult. The difficulty is reflected by the diversity in techniques available to assess the brain at early age. The techniques vary from clinical bed-side methods requiring no equipment, such as the various forms of neurological assessment, to more or less sophisticated technical assessments, such as brain imaging (ultrasound, magnetic resonance imaging and computer tomography) and neurophysiological tests, including EEG-recordings and visual or somatosensory evoked potentials. The sensitivities, specificities and accu-

racies of all these assessment techniques to predict developmental outcome show a large variation (1). The heterogeneity in predictive validity points to the need for advanced and more accurately described methods. Relatively recently a new assessment technique was developed. It consists of the evaluation of the quality of spontaneous motility of the fetus and young infant. In the next paragraphs the strengths and limitations of this new technique, i. e. the assessment of the quality of general movements, are discussed.

Assessment of the quality of general movements

Heinz Prechtl, a pioneer in the field of early neurological development, recognized the significance of spontaneous motor behavior in early life. He realized that self-generated motility during early development plays an important role in survival and adaptation (2). In addition, Prechtl discovered that the quality of spontaneous motility, especially the

quality of general movements (GMs) accurately reflects the condition of the nervous system of the fetus and young infant (3).

Typical development of general movements

GMs consist of series of gross movements of variable speed and amplitude, which involve all parts of the body but lack a distinctive sequencing of the participating body parts (2). Remarkably, GMs are amongst the first movements which the human fetus develops. A recent, detailed ultrasound study on the emergence of fetal motility revealed that the earliest movements can be observed at the age of 7 weeks and 2 days postmenstrual age (PMA) (4). The first movements are slow, small sideways bending movements of head and/or trunk. A few days later, the simple and stereotyped movements develop into movements in which also one or two arms or legs participate. But the movements continue to be slow, small, simple and stereotyped. At the age of 9

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Table 1
Age-specific characteristics of normal GMs (10, 12)

Tablica 1.
Obilježja normalnih OP ovisno o dobi (10, 12)

GM-type	Period of presence in weeks PMA	Description
Preterm GMs	from ± 28 wk. till 36-38 wk.	extremely variable movements, including many pelvic tilts and trunk movements
Writhing GMs	from 36-38 wk. till 46-52 wk.	something forceful (writhing) has been to the variable movements. In comparison to preterm GMs, writhing GMs seem to be somewhat slower and to show less participation of the pelvis and trunk
Fidgety GMs	from 46-52 wk. till 54-58 wk.	Basic motility consists of a continuous flow of small and elegant movements occurring irregularly all over the body, i. e. head, trunk and limbs participate to a similar extent. The small movements can be superimposed by large and fast movements

At any GM-age, the basic characteristics of normal GMs are: participation of all body parts; and movement complexity and variation.

to 10 weeks PMA GMs - consisting of movements in which all parts of the body participate - emerge. With the emergence of GMs the relatively simple sideways bending movements disappear. Initially, GMs show little variation in movement direction, amplitude and speed. But after a few days, the majority of GMs show a substantial degree of variation in speed, amplitude, participating body parts and movement direction.

GMs continue to be present throughout pregnancy and during the first months after term age. The incidence of GMs is relatively stable till 28 to 32 weeks PMA (5, 6). Thereafter the incidence decreases - a decrease which has been observed in utero and in preterm infants (7, 8). It should however be stressed that throughout pre- and postnatal life the incidence of GMs is characterized by a large intra- and interindividual variation (6, 7, 9).

GMs show age-specific characteristics (Table 1). Little is known on the age-specific changes of GMs during the first two trimesters of pregnancy. During the third trimester GMs are characterized by a large variation and complexity. The movements - described as "preterm" GMs - give the impression of a wonderfully complex ballet performance and include many movements of the trunk (10). Around 36-38 weeks PMA a transition in GMs can be observed. The largely

variable "preterm" GMs change into the more slow and forceful "writhing" GMs, in which the trunk participates less obviously than during the previous GM-phase (10). The "writhing" GMs constitute a temporary form of GMs, as they disappear around 6-8 weeks post-term age. The "writhing" GMs are replaced by the final form of GMs, the so-called "fidgety" GMs. "Fidgety" GMs consist of a continuous stream of tiny, elegant movements occurring irregularly all over the body (11, 12). The "fidgety" GMs disappear around 4 months post term. They are gradually replaced by goal-directed movements, such as mutually manipulative finger movements and reaching movements (11, 12).

Recently the hypothesis has been advanced that GM-complexity and variation is brought about by the transiently present cortical subplate (13). Basic arguments for the hypothesis are that: complex and variable GM-activity and the prominent presence of the subplate are both typical human (or primate) developmental features; and the evolution of the subplate neatly matches that of GM-development.

Subplate activity emerges when GM-complexity and variation emerge. The subplate is particularly present when GMs bloom, i. e. between 24 and 36 weeks PMA (14, 15). The gradual disap-

pearance of the subplate after 36 weeks PMA parallels the final phases of GM-development.

Abnormal GMs

Characteristics of abnormal GMs

Keywords to describe the quality of GMs are variation and complexity (Figure 1) (3, 16, 17). Complexity points to the spatial variation of the movements. Complex movements are movements during which the infant actively produces frequent changes in direction of the participating body parts. The changes in movement direction are brought about by continuously varying combinations of flexion-extension, abduction-adduction and endorotation-exorotation of the participating joints. GM-variation represents the temporal variation of the movements. It means that the infant produces continuously new movement patterns across time. Thus, the primary parameters of GM-quality evaluate two aspects of movement variation. This fits to the idea that variation is a fundamental feature of the function of the healthy young nervous system and stereotypy a hall-mark of early brain dysfunction (18, 19).

Four classes of GM-quality can be distinguished: two forms of normal GMs (normal-optimal and normal-suboptimal GMs) and two forms of abnormal GMs (mildly and definitely abnormal GMs; Table 2). Normal-optimal GMs are abundantly variable and complex. In addition they are also fluent. Normal-optimal movements are relatively rare: only 10-20% of 3 months old term infants show GMs of such a beautiful quality (20). The majority of infants shows normal-suboptimal movements, which are sufficiently variable and complex but not fluent. Mildly abnormal GMs are insufficiently variable and complex and not fluent, and definitely abnormal GMs are virtually devoid of complexity, variation and fluency. It is good to realize, that the classification into four categories of quality is somewhat artificial.

In fact, quality of movement is a continuum with at the one extreme splendidly complex, variable and fluent

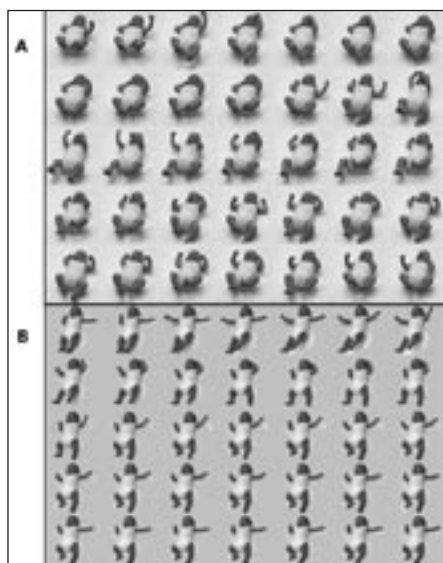


Figure 1
Representation of video-frames with GMs of two infants at fidgety-GM age. De video-recordings start in the left hand upper corner and should be read as the lines in a book. The interval between the video frames is 0,24 s. The infant in panel A was born at term and shows normal fidgety GMs. The continuously varying positions of the limbs illustrate the rich spatial and temporal variation of normal movements. The infant in panel B was born at 28 weeks PMA. She shows definitely abnormal GMs. The abnormal character of the movement is reflected by the lack of variation, indicated by the virtually identical frames, which induce the false impression that the infant hardly moves. (Video-recordings made in collaboration with the Dept. of Developmental and Experimental Clinical Psychology; figure published with permission of the parents and the Netherlands (37))

Slika 1.
Prikaz video zapisa s OP dvoje dojenčadi u dobi fidgety OP. Video zapisi počinju u lijevoj gornjem uglu i čitaju se poput crta u knjizi. Razmak između slika je 0,24 sekunde. Dojenče u zapisu A je došlo i pokazuje normalne fidgety OP. Stalna promjena položaja udova pokazuje bogatu prostornu i vremensku varijaciju normalnih pokreta. Dojenče u zapisu B rođeno je s 28 tjedana gestacije. Ono pokazuje definitivno abnormalne OP. Abnormalno obilježje pokreta očituje se nedostatkom varijabilnosti, na što upućuju slike skoro istog izgleda, što upućuje na lažni dojam da se dojenče jedva pokreće. (Video zapisi su urađeni u suradnji s Odjelom za razvojnu i eksperimentalnu kliničku psihologiju; slike su objavljene s dopuštenjem roditelja i Nizozemske (37))

movements and at the other extreme very stereotyped movements, such as a repertoire restricted to cramped-synchronized movements (10, 21). The latter movements are characterized by a suddenly occurring *en bloc* movement, in which trunk and - flexed or extended - limbs stiffly move in utter synchrony. Actually the cramped-synchronized movements are the only form of GMs which can be

Table 2
Classification of the quality of GMs (27)

Tablica 2.
Razvrstavanje kakvoće OP (27)

Classification	Complexity	Variation	Fluency
Normal-optimal GMs	+++	+++	+
Normal-suboptimal GMs	++	++	-
Mildly abnormal GMs	+	+	-
Definitely abnormal GMs	-	-	-

Complexity and variation: +++ = abundantly present, ++ = sufficiently present, + = present, but insufficiently, - = virtually absent or absent.

Fluency (the least important aspect of GM-assessment): + = present, - = absent.

considered as pathological. Their presence points to a loss of supraspinal control (22). Thus, the presence of cramped-synchronized GMs implies that the infant shows abnormal GMs. When an infant only occasionally shows a cramped-synchronized GM within a repertoire of movements which mostly exhibit some degree of variation and complexity, GM-quality can be classified as mildly abnormal. But when the infant frequently exhibits the cramped-synchronized pattern, GM-quality should be considered as definitely abnormal (23).

Validity of abnormal GMs

Various pre-, peri- and neonatal adversities, such as maternal diabetes, intrauterine growth retardation, preterm birth, perinatal asphyxia, neonatal hyperbilirubinaemia and neonatal treatment with dexamethasone can give rise to abnormal GMs (1). Definitely abnormal GMs are specifically but not exclusively related to discernible lesions of the brain (10, 21, 24). Recently evidence has been provided that definitely abnormal GMs are related in particular to periventricular white matter pathology, i. e. to damage of the efferent connections of the subplate. Whether injury to subplate neurons itself contributes to the generation of abnormal GMs is not clear (13).

It has also been demonstrated that movement quality is not a fixed phenomenon. It can change in various ways: movement quality can be transiently affected by illness and movement abnormalities can vanish or become more distinct with increasing age (25). The

majority of changes in GM-quality occur in the transitional periods during which normal GMs change in form, i. e., between 36-38 weeks PMA and between 6-8 weeks post term (26, 27). Within the three GM-phases (Table 1) movement quality is relatively stable.

The predictive validity of GM-quality varies with the age at which the GMs are evaluated and with the type of outcome. The best prediction can be obtained by longitudinal series of GM assessments. Infants who persistently show definitely abnormal GMs, even while passing the transformational phases at 36-38 weeks PMA and 6-8 weeks post term, have a high risk (70-85%) for the development of cerebral palsy (CP) (21, 24). Infants who persistently show cramped-synchronized GMs invariably develop CP (28). The prediction of a single GM assessment improves with increasing age. Thus, prediction is best at the age of fidgety GMs, i. e. at 2-4 months post term. Studies in populations of infants at very high risk for developmental disorders reported that the presence of definitely abnormal GMs at fidgety age, which implies a total absence of the elegant, dancing complexity of fidgety movements, predict CP with an accuracy of 85-98% (27, 29).

However, a current study on the development of preterm infants with less risk for developmental disorders than those included in the studies of Prechtl and Hadders-Algra indicates that the risk of the development of CP in infants with definitely abnormal GMs at "fidgety" age is substantially lower than about 90% and

lies in the order of 20-25% (30). Nevertheless, it should be realized that infants with definitely abnormal GMs at "fidgety" age who do not develop CP in general show other developmental problems, such as minor neurological dysfunction (MND), attention deficit hyperactivity disorder (ADHD) or cognitive problems (23, 27, 30). Mildly abnormal GMs at "fidgety" age are related to the development of MND, in particular to coordination problems and fine manipulative disability, ADHD and aggressive behavior, but the accuracy to predict these "minor" problems is modest, due to the presence of relatively many false positives, resulting in a moderate specificity (23, 26, 27, 31). The power to predict "minor" developmental disorders improves considerably when the results of the assessment of GMs are combined with those of the infant neurological examination (27).

Technique and reliability of GM-assessment

The assessment of the quality of GMs focuses on the amount of movement variation and complexity exhibited by the infant (Figure 1). These parameters can be appreciated by means of Gestalt perception of the observer (3). Gestalt perception allows the evaluation of the repertoire of movement patterns displayed by *all* parts of the body and does not pay special attention to particular behavior of specific body parts (e. g., fisting). GM-evaluation also includes the evaluation of movement fluency (Table 2). But this is the least important aspect of the assessment. Regrettably, our visual system has an innate sensitivity to spot a loss of movement fluency and this visual propensity for the detection of abnormalities in movement fluency, such as jerkiness, tremulousness and stiffness, interferes to some extent with the assessment of the major components of the GMs, i. e., movement complexity and variation.

The evaluation of movement complexity and variation is demanding and requires off-line assessment by means of a video-recording. Assessment of the movements in "real life" introduces errors and should be avoided (32). Ide-

ally, about 5 to 10 minutes of real-time motility is recorded with the infant in an adequate behavioral state. The absolute minimum duration of a GM-video is 3 minutes with real-time behavior. Only this minimum duration allows for an evaluation of the overall variation in the infant's motor repertoire. The video has the advantage that it also offers the opportunity of movement replay at high speed, which facilitates the evaluation of movement complexity and variation. A high speed replay produces an effect which is comparable to the effect produced by the video-frame sampling procedure of Figure 1.

GMs are affected by the behavioral state of the infant (33). The optimal state for GM-analysis is active wakefulness, i. e., Prechtl's state 4 (34). In this state the splendid variation and fluency of normal GMs is expressed best. During other behavioral states normal GMs get features reminiscent of abnormality, implying that a non-optimal state interferes with movement classification. When a video-recording only contains GMs during state 2 ("REM-sleep" or state-2-like conditions), the primary parameters of GM-analysis - complexity and variation - still can be evaluated. GMs should not be assessed during crying or non-nutritive sucking, including thumb sucking (33).

The basic principles of GM assessment can be learned in two days. Thereafter it requires further practice of about hundred GM-recordings to become a skilled observer (32). Various studies reported that the intra- and inter-observer agreement of GM-assessment of skilled observers is high (kappa-values around 0.80, implying an excellent interrater and intrarater reliability) (23, 26).

Concluding remarks

The assessment of the quality of GMs is a sensitive tool to evaluate brain function in young infants. It has a complementary function to the traditional neurological examination. Prediction of developmental outcome on the basis of longitudinal series of GM-assessment is best. Second best is prediction on the basis of an assessment at fidgety age. The

presence of definitely abnormal GMs at fidgety age puts a child at such a high risk for CP that it warrants physiotherapeutic intervention. It is unlikely that the intervention will prevent the development of CP, but animal data suggest that early intervention could improve the child's later functional abilities (35). Of course, this is an issue begging for further exploration and research, as up till now studies failed to prove a consistent positive effect of early intervention on long term motor development (36).

The clinical implications of the information that a child shows mildly abnormal GMs at fidgety age are less clear. It could be surmised, that mildly abnormal GMs point to minor developmental deviations in the cortical-subcortical circuitries, which put the infant at risk for the development of problems like MND and ADHD. However, the impact of the risk needs to be determined by future investigations in the general population.

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Sažetak

KAKVOĆA OPĆIH POKRETA KAO NAČIN PROCJENE INTEGRITETA MLADOG ŽIVČANOG SUSTAVA

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Prepoznavanje djece s razvojnim poremećajima, poput cerebralne paralize, u ranoj dobi je krajnje teško. Nedavno je razvijen novi način procjene mlade dojenčadi, temeljen na kakvoći općih pokreta (OP). OP su pokreti fetusa i malog dojenčeta u kojima sudjeluju svi dijelovi tijela. Obilježje tipičnih OP je složenost i varijabilnost pokreta; u abnormalnim OP složenost i varijabilnost je smanjena ili odsutna. Nedavno je iznijeta pretpostavka da je izvor složenost i varijabilnosti OP prolazna prisutnost subplate zone (zone pod pločom) i da su abnormalni OP posljedica oštećenja ili disfunkcije zone pod pločom i njenih referentnih motoričkih sveza u periventrikulskoj bijeloj tvari. Više istraživanja je ukazalo da abnormalni OP mogu predvidjeti razvojni ishod. Predviđanje temeljeno na longitudinalnom praćenju OP je najbolje. Drugo najbolje predviđanje je temeljeno na procjeni "fidgety" dobi OP (dob vrpoljenja) tj. u dobi od 2-4 mjeseca života. Definitivno abnormalni OP u fidgety dobi povezani su s cerebralnom paralizom, blago abnormalni OP sa minor neurološkom disfunkcijom u školskoj dobi. To upućuje da definitivno abnormalni OP u "fidgety" dobi ukazuju na potrebu rane fizioterapijske intervencije.

Deskriptori: OPĆI POKRETI, DOJENAŠTVO, PREDVIĐANJE, RANO ZAPAŽANJE, CEREBRALNA PARALIZA