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General Movement Assessment from videos of computed 3D infant body models is equally effective compared to conventional RGB video rating

A. Sebastian Schroeder^{a,d,1,2,*}, Nikolas Hesse^{b,1,2,3}, Raphael Weinberger^{a,e,1}, Uta Tacke^{a,c,1}, Lucia Gerstl^{a,1}, Anne Hilgendorff^{a,d,1}, Florian Heinen^{a,d,1}, Michael Arens^{h,1}, Linze J. Dijkstra^{f,1}, Sergi Pujades Rocamora^{i,1}, Michael J. Black^{g,1}, Christoph Bodensteiner^{h,1}, Mijna Hadders-Algra^{f,1}

^a Ludwig Maximilian University of Munich (LMU), Hauner Children's Hospital, Department of Paediatric Neurology and Developmental Medicine, Munich, Germany

^b Swiss Children's Rehab, University Children's Hospital Zurich, Affoltern am Albis, Switzerland

^c University Children's Hospital, Basel, Switzerland

^d Ludwig Maximilian University of Munich (LMU), Comprehensive Developmental Care (CDeC), Munich, Germany

^e Ludwig Maximilian University of Munich (LMU), Department of Epidemiology, Institute of Social Paediatrics and Adolescent Medicine, Munich, Germany

^f University of Groningen, University Medical Center Groningen, Department of Paediatrics, Groningen, Netherlands

^g Max Planck Institute for Intelligent Systems, Tübingen, Germany

^h Fraunhofer Institute of Optronics, System Technologies and Image Exploitation, Ettlingen, Germany

ⁱ Université Grenoble Alpes, Inria, CNRS, Grenoble INP, LJK, Grenoble, France.

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ABSTRACT

Background: General Movement Assessment (GMA) is a powerful tool to predict Cerebral Palsy (CP). Yet, GMA requires substantial training challenging its broad implementation in clinical routine. This inspired a world-wide quest for automated GMA.

Aims: To test whether a low-cost, marker-less system for three-dimensional motion capture from RGB depth sequences using a whole body infant model may serve as the basis for automated GMA.

Study design: Clinical case study at an academic neurodevelopmental outpatient clinic.

Subjects: Twenty-nine high risk infants were assessed at their clinical follow-up at 2–4 month corrected age (CA). Their neurodevelopmental outcome was assessed regularly up to 12–31 months CA.

Outcome measures: GMA according to Hadders-Algra by a masked GMA-expert of conventional and computed 3D body model ("SMIL motion") videos of the same GMs. Agreement between both GMAs was tested using dichotomous and graded scaling with Kappa and intraclass correlations, respectively. Sensitivity and specificity to predict CP at ≥ 12 months CA were assessed.

Results: Agreement of the two GMA ratings was moderate-good for GM-complexity ($\kappa = 0.58$; ICC = 0.874 [95%CI 0.730; 0.941]) and substantial-good for fidgety movements (FMs; Kappa = 0.78, ICC = 0.926 [95%CI 0.843; 0.965]). Five children were diagnosed with CP (four bilateral, one unilateral CP). The GMs of the child with unilateral CP were twice rated as mildly abnormal with FMs. GM-complexity and somewhat less FMs, of both conventional and SMIL motion videos predicted bilateral CP comparably to published literature.

Conclusions: Our computed infant 3D full body model is an attractive starting point for automated GMA in infants at risk of CP.

Abbreviations: BSID, Bayley Scales of Infant Development; CA, corrected age; CP, cerebral palsy; DA, definitely abnormal GMA rating; FMs, fidgety movements; GA, gestational age; GMA, General Movement Assessment; GMs, general movements; MA, mildly abnormal GMA rating; NS, normal suboptimal GMA rating; NO, normal optimal GMA rating; SMIL, Skinned Multi-Infant Linear Model; SMPL, Skinned Multi-Person Linear Model; RGB-D, Red Green Blue-Depth

* Corresponding author at: Ludwig Maximilian University of Munich (LMU), Hauner Children's Hospital, Paediatric Neurology, Developmental Medicine, Lindwurmstr. 4, 80337 Munich, Germany.

E-mail address: Sebastian.Schroeder@med.lmu.de (A.S. Schroeder).

¹ All authors have approved the final article.

² Authors contributed equally.

³ Work was done while NH was with Fraunhofer Institute of Optronics, System Technologies and Image Exploitation, Ettlingen, Germany.

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Cerebral palsy (CP) describes the most common physical disability in children. It occurs in approximately 2/1,000 live births [1]. The average age at which the diagnosis is made and communicated with the families has been shown to be approximately 11 months. For less severely affected children diagnosis may even be made as late as 24 months of age [2]. Yet, early and accurate detection of infants at very high risk of CP is a prerequisite for timely initiation of early intervention, i.e. intervention starting before the age of 6 months [3]. Early intervention programs use caregiver coaching and address the infant's sensorimotor development, attention, self-regulation, early communication skills and parental mental well-being [4,5].

To detect very high risk of CP, it is recommended to use a combination of standardized tools in conjunction with clinical history. Before 5 months corrected age (CA), the most predictive tools to detect risk of CP are term-age cerebral magnetic resonance imaging (MRI) and the video-based Qualitative Assessment of General Movements (GMA) between 2 and 5 months CA [6–8]. Term-age MRI may, however, not serve as a screening tool in clinical practice, leaving GMA as the most predictive tool in everyday practice.

General movements (GMs) are spontaneous movements, which involve all body parts [9,10]. They emerge during early fetal life and disappear when goal-directed motor behaviour emerges around 4–5 months CA. The form of typical GMs changes as a result of developmental transformations of the nervous system. In the last phase, at 2 to 5 months CA, GMs have a 'fidgety' character. Fidgety GMs (FMs) occur irregularly all over the body and consist of a continuous stream of tiny elegant movements. During each phase, typical GMs are primarily characterized by complexity and variation (in short: complexity) [10]. GMs are considered as abnormal when their complexity is reduced. [9,10]. At fidgety age – the age at which GMA best predicts developmental outcome [6,11] – atypical GMs are also characterized by a reduction or disappearance of the age-specific FMs. Therefore Hadders-Algra suggested that GMA at 2–5 months CA (fidgety age) is performed with a two-step procedure: (1) grading movement complexity and (2) assessing FMs [11,12].

Unfortunately, GMA requires a high level of expertise in the raters, which impedes the application in broad clinical practice [13–17]. Therefore, it is desirable to develop an easy to use, automated screening tool that can be applied in general paediatric practice as an alternative for the expert dependent video ratings of GMs. To be generally applicable, this diagnostic tool should be sensitive and specific and should use low-cost, commercially available equipment that does not influence the infants' motions.

A primary prerequisite for automatic analysis of motor development is to capture infant movements with high precision. Motions are represented as continuous time-series of the kinematics of limb and trunk movements, e.g., 2-dimensional (2D) or 3-dimensional (3D) body joint positions or joint angles. In the subsequent analysis stage, motion features have to be extracted from the captured movements to serve as input to machine learning classifiers, which can be used to identify infants with abnormal motor activity or to predict the risk of CP.

The first computerized approach towards automated detection of CP used a marker-based Vicon system to track movements in 3D [18]. Relevant characteristics of the captured movements were encoded in motion features based on limb velocities and accelerations. A machine learning classifier was trained to predict the CP risk based on these features [18]. Up to date, a wide variety of sensors with different properties have been used for capturing infant movements, e.g., body-worn miniaturized movement sensors [19,20] or magnetic tracking systems [21]. Video-based motion tracking has the advantage that it does not require the attachment of sensors [22,23].

Marschik et al. [24] developed the 'iDN Fingerprint Model' that combines multiple biological signals of the young infant, including a 3D video of GMs with a Red-Green-Blue Depth (RGB-D) camera. The Italian group of Orlandi and colleagues proposed a system based on the combination of audio and video data, i.e., the infant's spontaneous

vocalizations and its GMs [25]. However, currently both systems' psychometric properties have not been reported.

Purely computer-based 2D video analysis for the assessment of GMs was first published by Adde and colleagues [26]. They developed a General Movement Toolbox (GMT), which uses the displacement of pixels from one video frame to the next for quantitative analyses of GMs occurring at 2–5 months CA, i.e., the fidgety GMs. Visual representations from GMT resulted in recognisable patterns of the fidgety movements. They reported that the application of the automatic movement classification in their mixed study group of 82 high and low risk infants born at term or preterm would reduce the need for further referral by 70%. Their study showed that video recordings can be used for qualitative and quantitative analyses of GMs [26]. More recently, the same group suggested on the basis of a group of 150 high risk infants that the analysis could be used to reduce the rate of infants needing intensive developmental monitoring [27]. However, two things should be noted. First, the system misclassified some children, e.g., missing those who needed monitoring (10%) or referring those who did not require intensive follow-up (20%). Second, the authors had to exclude infants with a specific type of moderately abnormal GMs (abnormal FMs) from the analysis as the algorithm could not deal with these movements.

More recently, two other groups reported on computer-based analysis of 2D GM-videos. The group of Orlandi used an approach based on a skin model for segmentation and large displacement optical flow (LDOF) [28]. The kinematic features extracted from the model were used to classify fidgety GMs as typical or atypical. Their retrospective study based on 127 videos of preterm infants showed a specificity of 99%, but a sensitivity of only 44% in predicting CP [28]. The group of Marchi and colleagues reported an approach based on automated pose estimation to capture key aspects of GMs [29]. They used OpenPose - an open source computer vision software - developed by Cao et al. [30] to analyse the 2D videos of 21 preselected infants at fidgety GM-age. They considered their approach as feasible for automatizing infant motion analysis. However, they indicated that pose tracking errors occurred in 14 out of 21 recorded videos, which hindered automatized analysis. They concluded, that their approach would improve significantly by the use of three dimensional (3D) instead of 2D camera technology [29].

To summarize, up to now no results are available on the application of 3D systems for automated assessment of movements in infancy that do not use sensors/markers that have to be attached to the infants' limbs, which may affect their behaviour.

Since 2015, we have developed a motion tracking system that estimates the full body pose of infants in 3D [31–34] (Kinematic Motion Analysis Tool; KineMAT). The system is based on a commercially available, low-cost RGB-depth sensor (Kinect 1.0, Microsoft, USA) and can be applied without attaching markers to the infant's body (see Fig. 1) [31–33]. Recently, we developed the Skinned Multi-Infant Linear (SMIL) body model [34], which is based on the adult body model SMPL (Skinned Multi-Person Linear Model) [35]. The pose and shape of this virtual body can be adjusted with the model parameters. This allows us to use SMIL for capturing the 3D shape and 3D pose of infants from RGB-D sequences. We create a video of the resulting SMIL body reproducing the movements of the recorded infant, which we denote "SMIL motion video". Fig. 2 illustrates the resemblance of the RGB and SMIL motion videos.

The aim of this study was to evaluate how reliably our developed 3D infant body model (SMIL motion video) represents true infant spontaneous movements to a human observer. To this end, we assessed the agreement on the classifications of a human expert of GM-quality of two videos of the same GMs of 29 consecutively recruited high risk infants at fidgety age. The data of both videos originated from the same camera recording. One video consisted of the conventional GM-video used in clinical practice, the other of the SMIL motion video (Figs. 1 and 2). Both videos were assessed according to Hadders-Algra, i.e., assessing GM-complexity (in the present study with three degrees of fine-grading) and assessing FMs with two degrees of grading [12,36,37].

Fig. 1. Study setup.

We recorded General Movements of infants in the clinical setting of an outpatient clinic. We correlated expert GMA ratings of standard RGB videos with GMA ratings on SMIL motion videos of the same sequence. SMIL motion videos are the result of capturing 3D shape and motion with the SMIL model [34] (further explanation see: <https://www.youtube.com/watch?v=aahF1xGurmM&feature=youtu.be>).

1. Material and methods

1.1. Participants

Participants were high risk infants who received standardized follow up at the Centre of Developmental Care of the integrated Social Paediatric Centre at the University Hospital in Munich, Germany. High risk infants were infants meeting at least one of the following criteria: born ≤ 32 weeks of gestation (GA), birth weight below 1500 g, or other perinatal complications such as term asphyxia or early onset meningitis. Exclusion criteria were additional, potentially confounding diagnoses (e.g. genetic disorders, brachial plexus palsy, neuromuscular disorders). All infants meeting the inclusion criteria were examined by the first author between November 2016 and October 2017. The video recording was performed prior to the general developmental neurological examination. The infants' prenatal, perinatal and neonatal history was documented on standardized charts. After fidgety GM-age, the infants generally were assessed at a three to six months interval depending on the clinical need. Each follow-up visit consisted of a detailed neuropaediatric clinical examination. In children ≥ 12 months CA the diagnosis of CP was based on clinical findings in line with international recommendations as published by Boychuk et al. [38]. At 24 months CA, children were invited to participate in a structured neurodevelopmental assessment using the Bayley Scales of Infant and Toddler Development (BSITD, Bayley III Scales, full version).

The local ethics committee gave clearance for this study prior to the inclusion of the first patient (Ethikkommission LMU, Project Nr. 454-16). Parents gave written informed consent prior to study entry. No family withdrew consent during the study period.

1.2. Methods

The infant lay in supine position and an RGB-D camera (Microsoft Kinect V1, USA) was positioned directly above the infant at a distance of 100 cm. This camera allows simultaneous recordings of regular RGB videos (typically used for clinical GMA ratings) and depth videos, which provide the 3D distance to the camera for each pixel of the image. The infant's GMs were recorded for 3 min in a peaceful, quiet environment during active wakefulness.

To capture the infant's motion, we use SMIL (Skinned Multi-Infant Linear body model; see introduction for additional details on the development of the model [34]), which models the 3D body surface together with an underlying skeleton. The pose and shape of this virtual body can be adjusted with the model parameters. This allows the "registration" of the model to RGB-D data. Registration denotes the process of (automatically) adjusting model parameters so that the model best describes the input data, i.e. the model takes the same shape and pose as the recorded infant. We create a video of the SMIL registration results, i.e., the SMIL body reproducing the recorded infant movements, which we denote as "SMIL motion video" (see Figs. 1 and 2). SMIL allows access to 3D positions of all points of the body and the skeleton, as well as body joint angles in all degrees of freedom across time. The SMIL Model achieves a metric accuracy, i.e., the average distance of RGB-D scan points to the model surface, of 2.51 mm (SD 0.21 mm) and represents the original (correct) infant body pose in 98.8% of the time [34]. The model is adapted to the specifics of infantile body proportions. Our registration method can handle fast movements and self-occlusions, which are a common problem when capturing motions of

Fig. 2. Conventional video recording and its corresponding SMIL motion video pose.

The upper panel shows 8 representative poses of a three minute video recording of an infant at fidgety age. The infant presents with typical motion features of complexity and variation during active wakefulness; his GMA complexity was rated as normal suboptimal (Likert-score 7). The lower panel shows the corresponding SMIL motion video frames. In both panels GM-complexity can be clearly observed. Note that fidgety movements cannot be shown in the static poses of the figure.

freely moving humans with only one camera. Further details on the registration of the SMIL Model to RGB-D sequences can be found in previous publication [34].

The conventional video and the SMIL motion video of each infant were used for blinded GMA rating according to Hadders-Algra. For GMA at fidgety age this means a two-step procedure. First, the degree of GM-complexity is assessed – this is the parameter that can be assessed at any GM-age [10]. It supplies information on the integrity of the subcortical-cortical networks [11]. We graded GM-complexity in three ways: (a) by using a classification into four categories: normal-optimal movements (NO; abundant complexity and variation), normal-suboptimal movements (NS; sufficient complexity and variation), mildly abnormal movements (MA; insufficient variation and complexity; reflecting normal but non-optimal brain function) and definitely abnormal movements (DA; very limited complexity and variation including cramped synchronized movements; reflecting brain dysfunction) [10] (b) by applying a fine-graded 10-point Likert-scale, in which score 1 denotes a virtual absence of GM-complexity and 10 a very abundant GM-complexity [37]. Scores of NO movements ranged from 10 to 8, those of NS movements from 7 to 6, those of MA movements from 5 to 4 and those of DA movements from 3 to 1. The Likert-scale fine grading has been applied before; it helps to describe movement quality but it does not assist improvement of prediction of developmental outcome compared to that based on the four classes [37]. (c) GMA ratings were dichotomized into the clinically relevant DA category (1–3 on the Likert scale; high predictive value for CP) versus a non-DA category (MA, NS, NO, i.e. 4–10 on the Likert scale) as previously described by Hadders-Algra et al. [39]. Second, the degree of FMs is assessed: continuously present, intermittently present, sporadic and absent [40]. Continuously and intermittently present FMs were considered as typical FMs. In the calculation of predictive values FMs were categorized as absent or present, i.e., sporadic or typical. [12].

The senior author assessed all SMIL motion videos in random order (both in terms of GM-complexity and in terms of presence of FMs), followed by the assessment of all RGB videos in random order. The senior author is a GMA expert with >25 years of experience in clinical and scientific GMA rating (MH-A). She was masked for the infant's medical history and outcome.

1.3. Statistical analysis

Descriptive statistics were used to describe the infants' clinical information on single subject level (gestational age at birth, APGAR, umbilical cord pH and base excess, age at GMA recording, neonatal morbidity diagnoses, brain ultrasound findings, results of neurodevelopmental outcome at ≥ 12 months, GMA scores).

For the assessment of both videos by the same GMA rater, statistical tests representing test-retest and intra-rater reliability measures were used. The Intraclass Correlation Coefficient (ICC; two way mixed, absolute agreement) was used to evaluate the degree of agreement

between the two GM-ratings of GM-complexity using the 1–10 Likert scale and the FM-ratings. Reliability is considered high when ICC is 0.70 or higher. The ICC values are presented with 95% confidence intervals (CI). In addition, we used Bland-Altman plots to report the agreement between the GM-complexity on the basis of both videos.

Kappa statistics were used to describe the agreement of DA and Not-DA as well as for the classification of absent and present FMs of both types of video. Kappa-values of 0.41–0.60 are considered a “moderate agreement”, 0.61–0.80 a “substantial agreement” and values of 0.81–1.00 as “almost perfect agreement”. Two-by-two tables were used to describe the consistency of GMA for the conventional videos compared to those based on the SMIL motion video with respect to neurodevelopmental long-term outcome (CP present or absent). In addition, specificity, sensitivity, negative predictive value, and positive predictive value were calculated. Statistics were performed with IBM SPSS, version 25.0.0.1 (IBM Corporation USA). ROC curves were used to graphically illustrate the relationship between sensitivity and the occurrence of false positive values for DA versus non-DA GM-complexity ratings of both types of video.

2. Results

Twenty-nine infants met the inclusion criteria. Thus, 2×29 sets of videos were assessed. The infants' mean corrected age at recording was 14.8 weeks (95%CI 14.1;15.5). More than half (18/29, 62%) of the infants was born before 32 weeks of gestation. All infants born at higher gestational ages were high risk due to perinatal or postnatal complications (e.g. term asphyxia or meningitis) requiring standardized neurodevelopmental follow up examinations. At neurodevelopmental follow-up between 12 and 31 months CA 10 children (34%) showed a typical development; 14 had a global developmental delay (48%) and five (17%) had an ascertained diagnosis of CP (four a bilateral spastic CP, one a unilateral spastic CP). Detailed clinical characteristics of the infants are provided in Table 1 and Supplementary Table S1.

2.1. GMA rating: conventional video versus SMIL motion video

On the basis of GM-complexity assessments six infants were rated as DA, both in the conventional videos and in the SMIL motion video rating, 18 (13 SMIL motion video) as MA, and 6 (10) as NS. In both video ratings no infant was rated as NO (Table 1).

Using the dichotomy DA versus non-DA GM-complexity, 25/29 (86%) of both ratings were identical. The corresponding inter-rater agreement measured with the Cohen's Kappa was 0.58 ($p = 0.002$), meaning “moderate” agreement. For FMs present versus absent Cohen's Kappa was 0.78 ($p = 0.001$), meaning “substantial” agreement.

The GM-complexity ratings of both video methods using the 1–10 Likert scale were identical for 16/29 (55%) infants. An additional 10/

29 (34%) ratings showed only a ± 1 difference between both video-methods. The ratings of three infants (10%) differed more than one point. In two infants the ratings differed by two points (#6: conventional GMA rating of 5, corresponding to MA GM-complexity, and a SMIL motion video GMA rating of 3, corresponding to DA GM-complexity; #15 conventional GMA rating of 3 and a SMIL motion video GMA rating of 5). In the third infant the ratings differed by three points (#3, conventional GM-complexity 5 versus SMIL motion video GM-complexity 2) (Table 1, Bland-Altman-Plot see Supplementary Fig. S1). The mean Intraclass Correlation Coefficient (ICC) of both GM-complexity ratings was excellent 0.874 (95% CI 0.730;0.941).

In the FM rating of the conventional video FM's were absent in three infants (#1, #2, #6) of which two infants had DA GM-complexity and one MA GM-complexity. All three were later diagnosed with CP. In the SMIL motion video also three infants were rated as absent FM (#1, #6, #8) of which again two infants were rated having DA GM-complexity and one as having MA GM-complexity. Two of these were diagnosed with CP and one with global developmental delay at follow up. ICC of FM ratings was 0.926 (95% CI 0.843;0.965).

One infant who was diagnosed with bilateral CP (#5) and the infant with unilateral CP (#20) had presented either with sporadic or typical FMs in both videos (Table 1).

2.2. Prediction of developmental outcome

Table 2 summarizes the predictive values of the various GMA ratings including positive and negative predictive values. Sensitivity and specificity of DA vs non-DA rating of GM-complexity on the basis of the conventional videos to predict CP were 0.600 and 0.875, respectively; those of the corresponding SMIL motion videos 0.800 and 0.917,

respectively. The difference between the two is illustrated by ROC curves (Supplementary Fig. S2). Sensitivity and specificity of absent FMs of the conventional video were 0.600 and 1.000, those of the SMIL motion videos 0.400 and 1.000, respectively. Finally, the combination of GM-complexity and FM ratings resulted in a sensitivity and specificity of both types of video of 0.400 and 1.000, respectively. Child #20, later diagnosed with unilateral CP, was consistently missed: she showed MA GMs with FMs.

3. Discussion

Our clinical case-study indicated that GMA based on computer generated virtual 3D infant body models (SMIL motion videos) closely corresponds to the established gold standard based on conventional RGB videos. The SMIL motion videos were able to catch movement complexity and FMs. In addition, our data suggest that GMA ratings based on SMIL motion videos result in a similar prediction of CP as GMA based on conventional videos.

3.1. Comparison of GMA based on conventional versus SMIL motion videos

Several studies have reported excellent inter-observer agreement of skilled observers for conventional GMA [10,41,42]. Our comparison of the 29 simultaneously recorded video pairs could be considered a similar type of test-retest analysis. Corresponding to literature data, we found substantial inter-observer reliability (dichotomous FMs and GM-complexity Cohen's Kappa 0.78 and 0.58 respectively, finer graded ratings ICC 0.926 and 0.874 respectively).

The predictive values of both ratings of GM-complexity were comparable to those described in the literature; those of FMs were

Table 1
GMA Conventional video versus SMIL motion video.

Pat ID	sex	GA	CA	Clinical Diagnosis at 12–30 mo CA [38]	GM-complexity Likert score Conventional video	GM-complexity Likert score SMIL motion video	Fidgety movements Conventional video	Fidgety movements SMIL motion video
1	M	29 + 4	12	Bilateral CP	2	2	0	0
2	M	40 + 0	17	Bilateral CP	2	2	0	1
3	F	28 + 4	12	Dev. Delay	5	2	2	1
4	M	29 + 1	18	Dev. Delay	3	3	2	2
5	M	40 + 2	9	Bilateral CP	3	3	1	2
6	F	32 + 2	14	Bilateral CP	5	3	0	0
7	F	24 + 5	12	Dev. Delay	3	4	2	2
8	M	24 + 0	13	Dev. Delay	4	4	2	2
9	M	35 + 2	18	Age adequate	4	4	2	2
10	F	28 + 0	15	Dev. Delay	4	4	2	2
11	M	40 + 6	17	Age adequate	4	4	2	2
12	M	25 + 5	14	Dev. Delay	4	4	2	2
13	F	28 + 5	16	Age adequate	4	4	2	2
14	F	28 + 0	15	Dev. Delay	5	4	2	2
15	M	41 + 5	9	Age adequate	3	5	1	1
16	M	27 + 1	14	Dev. Delay	5	4	2	2
17	M	31 + 1	17	Age adequate	5	5	2	2
18	M	29 + 1	13	Age adequate	5	5	2	2
19	F	33 + 4	12	Age adequate	5	6	2	2
20	F	28 + 0	15	Unilateral CP	5	6	2	2
21	F	28 + 5	16	Age adequate	5	6	2	2
22	M	29 + 1	13	Age adequate	5	6	2	2
23	M	23 + 6	15	Dev. Delay	5	6	2	2
24	F	29 + 4	16	Dev. Delay	6	5	2	2
25	M	29 + 4	16	Dev. Delay	6	6	2	2
26	F	36 + 3	12	Dev. Delay	7	6	2	2
27	F	26 + 6	17	Age adequate	7	7	2	2
28	M	25 + 4	15	Dev. Delay	7	7	2	2
29	M	41 + 1	16	Dev. Delay	7	7	2	2

GM-complexity Likert scale ratings correspond to the following classifications 1–3 “definitely abnormal”, 4–5 “mildly abnormal”, 6–7 “normal suboptimal”, 8–10 “normal optimal”. Fidgety movements (FMs): 0 = absent; 1 = sporadically present; 2 = intermittently or continuously present. Abbreviations: CA = corrected age; CP = cerebral palsy; definition of CP in accordance with international expert recommendations [38]; terminology of the Surveillance of Cerebral Palsy in Europe (SCPE; [56]). DD: developmental delay, either based on performance on the Bayley Scales of Infant and Toddler Development (BSITD, Version 3) or on the clinical examination (see Supplementary Table 1); GA: gestational age; F: female; M: male; Bold: children with bilateral or unilateral CP

Table 2

Prediction of CP at follow up by conventional video GMA rating and GMA rating based on SMIL motion videos.

a) based on GM_complexity

		Cerebral palsy	No CP	Total	Q criteria
Conventional video					Sensitivity 0.6
DA-compl.		3	3	6	Specificity 0.875
Not-DA		2*	21	23	PPV 0.5
	Total	5	24	29	NPV 0.913
SMIL motion video					Sensitivity 0.8
DA compl.		4	2	6	Specificity 0.917
Not-DA		1*	22	23	PPV 0.667
	Total	5	24	29	NPV 0.957

b) based on FMs

		Cerebral palsy	No CP	Total	Q criteria
Conventional video					Sensitivity 0.6
FMs absent		3	0	3	Specificity 1
FMs present		2*	24	26	PPV 1
	Total	5	24	29	NPV 0.923
SMIL motion video					Sensitivity 0.4
FMs absent		2	0	2	Specificity 1
FMs present		3*	24	27	PPV 1
	Total	5	24	29	NPV 0.889

c) based on GM_complexity and FMs

		Cerebral palsy	No CP	Total	Q criteria
Conventional video					Sensitivity 0.4
DA-compl., FMs absent		2	0	2	Specificity 1
Not-DA, FMs absent		1	0	1	PPV 1
DA-compl., FMs present		1	3	4	NPV 0.889
Not-DA, FMs present		1*	21	22	
	Total	5	24	29	
SMIL motion video					Sensitivity 0.4
DA-compl, FMs absent		2	0	2	Specificity 1
Not-DA, FMs absent		0	0	0	PPV 1
DA-compl, FMs present		2	2	4	NPV 0.889
Not-DA, FMs present		1*	22	23	
	Total	5	24	29	

Abbreviations: DA compl. = definitely abnormal complexity GM rating (Likert Scale 1–3); Not-DA = all other GM ratings (mildly abnormal, normal suboptimal; Likert scale 4–7). FMs absent = fidgety movements scored “0”, FMs present = all other fidgety movements (sporadic, intermittent, continuously) [36,37], SMIL Skinned Multi-Infant Linear Body Model [34]. * Including infant #20 later diagnosed with unilateral spastic CP.

somewhat lower than the values reported in the literature, where summary estimates of sensitivity and specificity of GMA based on FM assessment have been reported to be 98% [95% CI: 74;100] and 91% [95% CI: 83;93%] respectively [6]. Interestingly, the predictive values based on the SMIL motion video were not inferior to those based on the conventional videos. While GM-complexity rating seemed to be superior using the SMIL motion video (Sensitivity 0.8 versus 0.6; PPV 0.667 versus 0.5, NPV 0.957 versus 0.913), the conventional video seemed to be slightly superior regarding the assessment of FMs (Sensitivity 0.6 versus 0.4; NPV 0.923 versus 0.889). It is conceivable that the SMIL motion video is superior to conventional GMA rating of GM-complexity, as the SMIL motion video forces the observer to focus on the Gestalt perception of GM-quality, whereas in the conventional video the assessor may be distracted and/or biased by additional visible details, such as naso-gastric tube, oxygen supply, monitoring cables or details of facial expression. Regarding the assessment of FMs, however, the SMIL motion video at times contains slight pose inaccuracies due to noisy input data, which may be misinterpreted as very small amplitude FMs. This could explain that it was occasionally hard to distinguish sporadic and absent FMs on the basis of the SMIL videos due to insufficient data quality. This underpins the conclusion, that assessing

both parameters (GM-complexity and FMs) is relevant for an optimal clinical prediction.

To understand the differences in the GMA ratings based on the conventional and SMIL motion videos of infants #2, #3, #6, and #15 MH-A reviewed these videos. She noticed the following for the infants (#3 and #6) whose GM-complexity was rated better on the conventional video (MA) than on the SMIL video (DA): Infant #3 (developmental delay at follow-up) showed especially some complex movements in the distal joints (with FMs), apparently these movements made a less prominent impression on the SMIL video. Infant #6 who was later diagnosed with bilateral CP had shown on the conventional video a discrepancy in movement complexity between the upper and the lower part of the body, with the upper being better than the lower. This discrepancy was less striking on the SMIL video. Both videos were rated as absent FMs. Infant #15 (typical outcome at follow-up) had been assessed at 9 weeks CA. GM-complexity of the conventional video was rated as DA, due to prominent stereotyped movement sequences in the proximal joints; the distal joints showed some complexity. The latter was more prominently visible during the SMIL video, which was therefore rated as MA. Both videos were rated as sporadic FMs. It has been shown by others, that movement quality at 9 weeks CA may

improve to better qualities during the following weeks [43]. Presumably this happened also in this infant. Finally, the GMs of infant #2 (bilateral CP) were rated on both videos as DA. However, the FMs were scored as absent on the conventional video, but as sporadic on the SMIL video – the few FMs had been observed at the wrists only. The latter may also be due to the slight pose inaccuracies due to noisy input data of the SMIL Model suggesting the presence of sporadic FM. The differences in scores between conventional and SMIL videos could be due to such technical matters, but they also underline that GMA based on Gestalt perception may remain difficult even after years of experience; it stresses the need of automatic movement analysis following strict algorithms.

GMA was especially adequate in the prediction of bilateral CP. Yet, it missed the child who was diagnosed with unilateral CP. It has been reported before that GMA does not detect all children with unilateral CP [44–46]. The differential ability of GMA to predict bilateral and unilateral CP corresponds to the putative neural substrate of the quality of GMs. GM-quality is considered to reflect the integrity of extensive neural networks involving not only cortical areas, but also their connectivity with subcortical relay stations [7]. Indeed, evidence is accumulating that DA GMs are especially related to damage of the periventricular white matter [7,47]. In part of the children with unilateral CP the lesion is restricted to the cortical grey or does only affect part of the cross-roads running through the periventricular white matter (as in infant #20) and does not massively disrupt white matter connectivity, therewith not producing DA GMs. The detection of children with unilateral CP is also challenging due to the finding that fidgety movements are frequently present.

Two children showed DA GMs (#4 and #7) but were not diagnosed with CP. They both presented with “global developmental delay” at follow-up. Recent data indicate that the limitations in the infant's early motor repertoire do not only predict an increased risk of CP, but also an increased risk of cognitive impairment irrespective of the GMA method used for classification [48–50].

3.2. Strengths and limitations

The strength of the current study is that it is the first that compares GMA based on videos of computed 3D infant body models to GMA rating of conventional videos. Another strength is that the videos were rated by one masked and highly experienced GMA assessor using multiple grading strategies including explicit assessment of FMs.

The study also has limitations: the sample size was small and consisted of high risk infants only. However, this convenience sample might be considered as representative of many infant follow-up clinics and represented infants with normal, MA and DA GMs. It may also be considered that the duration of follow-up was limited, which may mean that some children with a developmental diagnosis may have been missed [37].

3.3. Current status and future perspective of GMA based on SMIL modelling

In contrast to previous approaches that capture a limited number of body joint positions based on 2D-video [27–29,51,52] or rely on markers/sensors attached to the baby [18,19,53,54], our method allows the extraction of angles of all body joints (3 degrees of freedom per joint), 3D positions of body joints and of arbitrary points on the body surface without the need of potentially distracting markers. Our method is robust to self-occlusions (e.g. hands and feet occluding large parts of the body) and fast motion. It has been proven to display the true whole body movements of the recorded infant in 98.8% of recorded video sequences, and to have a metric accuracy, i.e. the average distance of RGB-D scan points to the model surface, of 2.51 mm (SD 0.21 mm) [34].

In order to further improve the SMIL tracking, we plan to integrate means to detect and fix the rare occurrence of failure cases, as recently

published by Aristidou et al. [55]. In addition, we aim to apply our method to a larger patient population, not only to improve the detection of predetermined motion parameters (e.g. fidgety general movements, centroid of motion, cramped synchronized activity, or a reduction of segmental movements), but also to use machine learning algorithms to quantify the motion parameters which are assumed to drive the “Gestalt perception”.

3.4. Concluding remarks

Our case study of 29 high risk infants demonstrated that the amount of motion details captured by the SMIL motion video - based on a low-cost Kinect recording and the KineMAT tool - enables accurate GMA at fidgety age. This implies that the SMIL motion video adequately catches the movement characteristics needed for GMA of infants with movements ranging from a normal to a definitely abnormal quality, turning it into an attractive tool for automatic GMA.

Declaration of competing interest

MJB is a co-founder and investor in Meshcapade GmbH, which is commercializing infant shape and motion technology.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.earlhumdev.2020.104967>.

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