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Title:

The In-Motion-App for remote General Movement Assessment: A multi-site observational study.

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Abstract

Objectives To determine whether videos taken by parents of their infants' spontaneous movements were in accordance with required standards in the In-Motion-App, and whether the videos could be remotely scored by a trained General Movement Assessment (GMA) observer. Additionally, to assess the feasibility of using homed-based video recordings for automated tracking of spontaneous movements, and to examine parents' perceptions and experiences of taking videos in their homes.
Design The study was a multi-center prospective observational study.
Setting Parents/families of high-risk infants in tertiary care follow-up programs in Norway, Denmark and Belgium.
Methods Parents/families were asked to video record their baby in accordance with the In-Motion standards which were based on published GMA criteria and criteria covering lighting and stability of smartphone. Videos were evaluated as GMA "scorable" or "non-scorable" based on pre-defined criteria. The accuracy of a 7-point body tracker software was compared to manually annotated body key points. Parents were surveyed about the In-Motion-App information and clarity.
Participants The sample comprised 86 parents/families of high-risk infants.
Results The 86 parent/families returned 130 videos, and 121 (96%) of them were in accordance with the requirements for GMA assessment. The 7-point body tracker software detected more than 80% of body key point positions correctly. Most families found the instructions for filming their baby easy to follow, and more than 90% reported that they did not become more worried about their child's development through using the instructions.
Conclusions This study reveals that a short instructional video enabled parents to video record their infant's spontaneous movements in compliance with the standards required for remote GMA. Further, an accurate automated body point software detecting infant body landmarks in smartphone videos will facilitate clinical and research use soon. Home-based video recordings could be performed without worrying parents about their child's development.

Strengths and limitations of this study

- A cohort of families of high-risk infants frequently seen in NICU follow-up settings.
- In-Motion-App standards for remote General Movement Assessment (GMA) communicated through a simple and short animated video.
- Data from a motion tracking software on smartphone videos pioneering automatic and markerless infant motion capture.
- Study not designed to evaluate reasons for families not recording or returning videos.
- Study did not evaluate how markerless infant motion capture on smartphone videos can be used for prediction of CP outcome.

1. INTRODUCTION

Cerebral Palsy (CP) is the most common physical disability in childhood. Diagnosis is typically set between 12- and 24-months corrected age (1-3). Early developmental screening of high-risk infants to predict future neurological impairments is today a priority for clinicians and researchers, and most parents express interest in such neurodevelopmental screening (3, 4).

The General Movement Assessment (GMA) has been recommended in combination with magnetic resonance imaging (MRI) to achieve a CP diagnosis before 6 months corrected age in infants with newborn-detectable risk factors (3). Early detection of CP has the potential to improve the organization and resources used in follow-up screening at hospitals and reduce medical complications for children with CP. The fidgety type of general movements (GMs) observed at 9-20 weeks' corrected age has shown high predictive validity for later CP (5-8). Video recordings for GMA must follow requirements for infant state, position and clothing and are scored by certified and trained assessors (9). Such trained GMA observers also have the expertise needed to ensure that video recordings fulfill the requirements for a valid GMA (9).

Access to trained observers using the GMA in hospital-based follow-up programs is limited by geographical constraints and lack of GMA expertise (10). As health care and parents move into the digital age using smartphones to share videos via internet, opportunities to perform remote GMA have developed. Recently, the Baby Moves smartphone app was presented for remote GMA within research settings (10, 11). However, smartphone apps for health data capturing in clinical settings are rarely assessed and usability tested (12). To be feasible in a clinical follow-up setting, home-based video recordings must fulfill basic GMA requirements without the need for comprehensive parental training or guiding.

Video recordings by hand-held smartphones introduce movement artifacts in the camera. Computer-based methods for objective detection of infant GMs (13, 14) may be jeopardized by such artifacts. Our research group has recently presented a machine-learning model which predicted CP with high accuracy comparable to observational GMA (15). Important shortcomings of the method were the need for manual and time-consuming body point annotations, as well as the need for a stationary camera. Hence, an automated 7-point body tracker has been developed by our group and needs to be validated on recordings taken with hand-held cameras.

Provided that GMs can be assessed, and computer-based infant body point tracking can be performed on videos taken with a hand-held camera, it is possible to perform remote GMA as well as automated infant body point tracking for a computer-based model for the prediction of CP. Thus, the In-Motion instructional video has been developed so that parents can perform home-based videos with quality standards feasible for remote GMA and automated infant body point tracking. Feasibility of the In-Motion instructional video was assessed in a multi-site study including families of high-risk infants from Norway, Denmark and Belgium.

The main aims of the study are as follows: 1) To determine whether videos taken by parents with a hand-held camera were in accordance with the standards set in the instructional video, and whether the videos could be scored by a trained GMA observer. 2) To assess the accuracy of a 7-point body tracker software based on the same recordings. 3) To describe parents' perceptions of the instructional video and filming their baby in a home environment.

2. METHODS

Design

Multi-center prospective observational study.

Patient and public involvement

The study protocol including the parental survey content was developed and designed collaboratively with representatives from The Norwegian Cerebral Palsy organization and The Norwegian Premature Association.

Participants

Parents of infants admitted to one of five participating level III-IV Neonatal Infant Care Units (NICU) in Norway, Denmark and Belgium from 2017-2018 were consecutively recruited at referral to the hospital follow-up program before discharge from the NICU. Families were recruited based on willingness to participate and the infant being evaluated as at high-risk of CP. In Norway and Denmark inclusion criteria were (a) Birth weight (BW) ≤1000 g (extremely low birth weight (ELBW) and/or gestational age (GA) <28 (extremely low GA (ELGA)), (b) neonatal arterial ischemic stroke, (c) neonatal encephalopathy, (d) other significant risk factors. In Belgium, only infants with GA <32 weeks or with perinatal stroke were included.

Data collection procedure

Included participants were assisted by a research physiotherapist/pediatrician to download and install the In-Motion-App containing the instructional video from Google Play or iTunes. They could ask any questions about the app and how to manage the software. They got information about the time window for performing two separate video recordings between 12^{+1} - 13^{+6} and 14^{+1} - 17^{+6} weeks post term age (PTA). The time points were defined to ensure GM videos from the fidgety movement's period. If no videos were returned from the families before 17^{+6} weeks PTA, the local study coordinator contacted the family to ask the reason why they had not uploaded any videos. The app was linked to a secure online server hosted at St. Olavs Hospital in Trondheim, Norway, and was available for i-operating system (iOS) and Android. After the end of the second time window, the families were contacted by email with a link to an online Norwegian University of Science and Technology survey to collect information about their opinions using the app and the In-Motion instructional video.

The In-Motion-App and instructional video

The In-Motion instructional video was designed for parents to give basic insight into recording standards needed for GMA and lighting and stability of camera. It was made as a short animation with simple drawn sequences containing a minimum of text. The instructional video was deployed to the parents by downloading the In-Motion-App developed for this study, and videos could be uploaded to be remotely assessed by a trained GMA observer.

After downloading the app and getting basic information from the local study coordinator, parents logged into the app with a username and a password. They typed in the first name of their child and the expected date of delivery (due date). The In-Motion-App generated two separate time windows between 12^{+1} - 13^{+6} and 14^{+1} - 17^{+6} weeks PTA and visualized them in a graphical timeline to show when videos should be taken. A red dot illustrated todays date placed on the timeline, helping parents to plan when to perform the video recordings. In addition, a pop-up message reminded parents to prepare for videos a week before the beginning of each time window. The In-Motion-App was constructed in such a way that the video recording automatically stopped after 3 minutes and asked the parents whether to upload the video or not.

The instructional video was 2 minutes and 47 seconds long. Before taking videos, parents were told to look through the In-Motion instructional video which was available from the app menu. They could watch the video as many times as they wanted until they felt confident performing the recording. The main themes aimed at ensuring quality standards for remote GMA included: a) clothing of infant, b) surface/underlay for infant, c) lighting, d) state of infant, e) positioning and f) length of video. In addition, instructions were provided about how to keep the smartphone steady and ensure

that the whole infant body was observable in the video image. Examples of some of the In-Motion instructional video themes are shown in Figure 1. Parents were asked to consecutively upload videos to the server at St. Olavs Hospital in Norway.

Assessment of video quality for remote GMA

Videos were assessed by a certified GMA observer with respect to the following standards (9): *1) GMA standards:* active movements (not hypokinetic), supine position, correct state, adequate clothing (diaper or a onsie), no disturbances during recording. *2) Additional In-Motion standards:* adequate light, whole body visible, feet of parent visible in video (ensuring correct position of smartphone camera, see picture to the right in Figure 1) and camera stability. Based on these standards, a classification was made by the same certified GMA observer as either "GMA scorable" if all standard criteria were fulfilled or "GMA non-scorable" if one or more standard criteria were inadequate. In addition, it was documented whether the hand-held video had optimal stability, events of abrupt displacement, was predominantly unstable, whether an adequate underlay was used (firm, comfortable, large enough) and whether overall video image quality was sufficient (blurred/very blurred).

Assessment of General Movements

All videos classified as "GMA scorable" were consecutively assessed by one certified and experienced GMA observer that had passed advanced GMs courses under the General Movement Trust (LA). The observer had no knowledge about the infant's clinical history. According to Prechtl's method of assessment of general movements (9), fidgety movements were classified as continuous (FM++), intermittent (FM+), sporadic (FM+/-), abnormal (Fa) or absent (FM-).

In-Motion body point tracking

A subset of 66 videos (5493 video frames) recorded by the In-Motion-App was used for automatic infant motion tracking to detect the displacement of 7-body points (nose, thorax (center between shoulders), wrists, pelvis, and ankles). The infant motion tracker algorithm consists of a convolutional neural net trained on 14900 video frames on high-risk infants that had participated in another study from our group (8). For further technical details of the convolutional neural net, the reader is referred to Groos et al (16). All 7 body points in the videos used in this study were manually annotated and used as ground truth for automatic body point position evaluation. The performance of the infant motion tracker is reported as percentage of points within a circular area around the annotated body point for the 5493 test frames. The size of the circular area was set to 10% of the infant head size and was normalized to adjust for different scaling (i.e., video zoom) (Figure 3).

Survey

Parents' opinions about the In-Motion-App and instructional video were collected using SelectSurvey v.4 software (ClassApps Inc., www.classapps.com) in Norway, operated by Norwegian University of Science and Technology (NTNU). In Denmark, parents' opinions were collected using Smart Trails data management software (MEDEI ApS. www.medei.dk) and in Belgium the survey was collected by post. The survey was sent to the families by a link in an email within one week after the last video was returned. The survey questions were customised for the In-Motion-App based on a tool developed by Jin and Kim in 2015 (17) and a survey used in a similar study on the Baby Moves App (11). It contained questions based on forced-choice questions covering the themes; 1) In-Motion-App, 2) In-Motion standards for remote GMA and 3) parental worries, were a statement was made and the parents indicated agreement or disagreement with the statement on a 5-point scale (Appendix S1, supplementary information).

Data analysis

Data were analyzed using SPSS statistics version 26.0 (IBM SPSS Statistics, Chicago, IL). The data are presented as numbers with proportion (%) or mean with standard deviation (SD) and range. Differences between infants with returned and no returned videos were analyzed using the Mann-Whitney U Test for continuous variables and Chi-square test and Fisher's Exact test for dichotomous variables. The accuracy of the 7-point body tracker was presented as the distribution of body point detections relative to the manually annotated body points, where 10% of the infant head size was used as a threshold.

3. RESULTS

In total, 86 infants/families were recruited and 17 (19.8%) out of them did not submit any video (Figure 2), leaving 69 families with 130 videos for analysis.

Twenty-eight (32.6%) families were included from three different hospitals in Norway, 43 (50%) from one hospital in Copenhagen, Denmark and 15 (17.4%) from one hospital in Gent, Belgium. Infant/family characteristics are shown in table 1.

3.1 Video recordings

Two (1.5%) out of 130 videos were shorter than 3 minutes (1 minute and 12 seconds and 2 minutes and 17 seconds). The mean PTA at video recording was 14.5 weeks (SD 2.28, range 8.1-23.6 weeks).

Fifteen (11.7%) families returned one recording, 49 (71%) two recordings, 4 (5.8%) three recordings, and 1 (1.4%) six recordings. One-hundred and seventeen (90%) videos were returned within the expected time window between 12^{+1} - 17^{+6} weeks PTA. Two (1.5%) families returned 2 and 3 videos, respectively, which were all taken outside the time window (week 8, 10, and 21 and 23, respectively). Eleven families (8.5%) with videos from within the requested time window, had additional videos taken outside the expected time windows. Six videos from one family were taken at two different days; four videos in weeks 12 and two videos in week 14 PTA.

3.2 Remote General Movement Assessment

Among the six videos returned from one family, the first one from each of the two days was selected for GMA analysis. Exclusion of the remaining 4 videos and additional 4 videos excluded due to PTA outside of the age required for assessment of FMs gave a total of 122 videos available for quality assessment.

One hundred and twenty-one (99%) out of 122 videos which were returned within the required time window were classified as GMA scorable. The video that was non-scorable had infant in side lying position. Details about compliance to the In-Motion standards are shown in Table 2.

3.3 General Movement Assessment

Of the 121 videos classified as GMA scorable, 3 (2.4%) videos were classified with exaggerated (Fa), 3 (2.4%) with absent (FM-), and 7 (5.6%) with sporadic (FM+/-) FMs. Eighty-seven (69%) and 21 (16.7%) videos were classified with intermittent (FM+) and continuous (FM++) FMs, respectively.

3.4 Computer-based body point tracking

The proportion of correctly predicted left wrist key point from 5493 tested video images was 83.15%. Details of accuracy of 7 predicted body points and mean value for all points are shown in Figure 3.

3.5 Parent responses

Survey responses were received from 64 (92.8%) families of the 69 families who returned at least one video. Fifty-four (84.3%) of them observed the instructional video one or two times before filming their baby. No families returned a video without training on filming their baby first.

The majority of the survey respondents found the In-Motion-App easy to use. All respondents agreed or strongly agreed that it was easy to understand how to stand and hold the smartphone during the filming. Details about family responses are shown in Table 3. Fifty-seven (90.5%) families strongly disagreed, disagreed or neither disagreed nor agreed that they did become more worried about their child's development through using the In-Motion instructions.

DISCUSSION

In this study, more than 95% of families with high-risk infants filming their baby at home, returned at least one video that was in accordance with the In-Motion standards for remote GMA. Most families found the In-Motion-App easy to use and the instructions for filming easy to follow, and less than 10% of respondents became worried through using the In-Motion-App. Despite the use of hand-held smartphones introducing movement artifacts in the video image, our computer-based 7-point body tracker detected positions of the body points with high accuracy. To the best of our knowledge, this is the first automatic infant body point tracker that is tested on video recordings from hand-held smartphones.

This study has several strengths. First, it included families of high-risk infants frequently seen in NICU follow-up settings. We argue that this makes our findings robust and generalizable to comparable clinical settings. Second, the communication of In-Motion standards through a simple and short animated video, makes the instructions easily applicable to a broad range of different clinical settings. Furthermore, the accuracy of the 7-point body tracker software with the use of smartphone videos makes it pioneering in the field of automatic and markerless infant motion capture compared to other studies (18, 19). This facilitates further development of methods for early automated detection of CP based on smartphone videos. Finally, the design of the study using an experienced and certified GMA observer for evaluation of video quality and a survey with very high response rate, makes the study quality high and the results trustworthy.

There are also several limitations. First, almost 20% of the included families did not return any video. This study was not designed to evaluate reasons for not recording or returning videos. We can, therefore, only conclude on the quality of returned videos. Problems encountered by families who did not record or return any video need to be further explored. Our findings are in accordance with the study by Kwong et al (11), where 24% of families did not return any video using the Baby Moves App. These findings indicate that home-based video recordings for remote GMA is not a solution for all families and that it might be difficult in a clinical setting to know beforehand which families will return a video or not. Furthermore, 13 (19%) families returned one or several videos outside the required time window needed for a valid GMA. More than 90% of respondents found the reminders in the App helpful, but one fifth disagreed that the number of reminders were appropriate. This indicates a limitation in the design of the app reminders to prepare parents for video recordings at the correct time. This needs to be improved in a process involving the users.

Second, our study comprised five different hospital sites in three different countries, and information provided to families when downloading the App may have differed. Additionally, there is a risk that the research personnel could have given more information and assistance to families than will be common in an ordinary clinical setting. Our study setting could therefore be in slight contrast to an ordinary clinical setting, where adapted and flexible family information is needed due to

challenges in reduced participation in neurodevelopmental follow-up (20) and racial and socioeconomic differences in mobile health technology usage (21).

Third, even though almost 20% of the hand-held videos in our study were classified as predominantly unstable, the GMA expert considered the videos to be GMA scorable. There is a risk that more videos might have been classified as non-scorable by another GMA observer with less training and experience. To the best of our knowledge, there is only one study protocol planning to assess the predictive validity of GMA from videos taken with hand-held smartphones for CP outcome (10). Hence, further studies on the use of smartphones for GMA are needed.

Finally, our computer-based 7-point body tracker showed accurate estimations compared to manual annotations on the video image. However, further studies must explore how selection of body points, tracked body point accuracy and movement artifacts in camera will influence a machine-learning model for prediction of CP from smartphone video recordings.

This study facilitates and contributes to the use of smartphone technology for video recordings and remote GMA. Consequently, it will contribute to giving high-risk infants and their families equal access to GMA as an accurate method for early identification of CP, without geographical constraints. The use of early remote medical assessment will improve the organization and resources used in follow-up screening at hospitals and have the potential to reduce medical complications for children with CP due to early detection. A clinical feasible computer-based movement analysis with equal accuracy as GMA, will greatly reduce the need for specialized GMA observers and provide an innovative resource-effective diagnostic measure.

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Contributors

Conceptualization; LA, RS, HP, OMS, TF, SO, WS, BE, CB, Data curation; LA, RS, EI, DG, Formal analysis; LA, RS, EI, DG, Funding acquisition; LA, RS, Methodology; LA, RS, EI, DG, TF, SO, Project administration; LA, BE, HP, AP, AB, CB, RS, Resources; LA, TF, SO, AB, AP, HP, OMS, BE, WS, Writing-original draft; LA, RS, EI, DG, Writing-review & editing; LA, RS, EI, DG, TF, SO, BE, WS, AB, AP, CB, HP, OMS.

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Competing interests

None declared.

Patient consent

Obtained in all participants.

Ethics approval

All infant/families provided written informed consent and ethics was approved by the regional committee for medical and health research ethics (REC Central-Committee 2017/913) in Norway. The

study sites in Denmark and Belgium also had approvals from their local Institutional Review boards. Study registration in ClinicalTrials.gov Protocol Record 2017/913.

Provenance and peer review
Not commissioned; external peer reviewed.

Data availability statement
No data are available. Data from this study are not available for sharing due to ethical approval requirements. Researchers interested in collaboration should contact the corresponding author with their expression of interest.

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Table 1 Summary of infant/family characteristics

	Total N (%) (*n*=86)	N (%) video responders (*n*=69)	N (%) no video responders (*n*=17)	*p*-value
Demographics				
Boys, n (%)	51(59.3)	42(60.9)	9(52.9)	0.32
BW, mean (SD), grams	1952(1107)	1915(1124)	2105(1055)	0.67
GA, mean (SD), weeks	32.4(5.3)	32.3(5.4)	33.4(4.7)	0.43
Risk group				
Birth weight (BW) ≤1000 g and/or gestational age (GA) <28	25(29.1)	20(29)	5(29.4)	0.77
Neonatal arterial ischemic stroke	11(12.8)	4(5.8)	7(41.2)	0.001
Hypoxic ischemic encephalopathy	20(23.3)	17(24.6)	3(17.6)	0.06
Others	32(37.2)	27(39.1)	5(29.4)	0.38
		Infant families (*n*=63)		
Socio-demographic data*				
Mother relation (survey), n (%)		48(76.2)		
Married/cohabitant family, n (%)		59(93.7)		
Age mother/farther, mean (SD, range)		31.8(5.5,21-6)		
Age farther, mean (SD, range)		33.9 (6.9, 22-59)		
Single child, n (%)		32 (51.6)		
iOS vs. Android				
iOS users, n (%)		41(65.1)		

Table 2 Compliance to In-Motion standards (n= 126)

	Active movements (not hypokinetic)	Supine position	Correct state	No disturbances during recording	Adequate clothing	Adequate light	Whole body visible	Feet of parents visible
N (%)	126 (100)	125 (99.2)	122 (96.8)	124 (98.4)	125 (99.2)	124 (98.4)	124 (98.4)	116 (92.1)*
		Optimal stability	Abrupt displacement	Predominantly unstable	Correct base of support		Image quality	
						Clear	Blurred	Very blurred
N (%)		80 (63.5)	26 (20.6)	22 (17.5)	119 (94.4)	114 (90.5)	11 (8.7)	1 (0.8)

*3 missing data.

Table 3 Parents' responses to the In-Motion-App and instructional video

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree
In-Motion_App					
The In-Motion app was generally easy to use	58,7%	33,3%	3,2%	4,8%	0,0%
It was easy to enter the information needed in the In-Motion app	46.0%	41.3%	11.1%	1.6%	0.0%
The reminders about when the child should be filmed were helpful	44.4%	41.3%	6.3%	6.3%	1.6%
The number of reminders about when the child should be filmed was suitable	25.4%	39.7%	15.9%	14.3%	4.8%
There were no technical problems with uploading and sending the videos	55.6%	22.2%	6.3%	4.8%	11.1%
In-Motion standards for remote GMA					
It was easy to understand how I should stand and hold the telephone during the filming	60.0%	40.0%	0.0%	0.0%	0.0%
It was easy to keep the telephone still while I was filming	16.7%	46.7%	30.0%	6.7%	0.0%
It was easy to do the filming without disturbing the child	36.5%	42.9%	9.5%	11.1%	0.0%
It was easy to understand how my child should be dressed when filmed	65.1%	33.3%	0.0%	1.6%	0.0%
It was easy to understand how my child should be positioned and how the mat should be when I was going to film	63.5%	28.6%	4.8%	3.2%	0.0%
It was easy to follow the instructions about how the lighting should be during the filming	36.5%	38.1%	14.3%	11.1%	0.0%
Filming my child for 3 minutes went smoothly	39.7%	39.7%	19.0%	1.6%	0.0%
Parental worries					
I felt safe about uploading video of my child	63.5%	27.0%	7.9%	1.6%	0.0%
I became more worried about my child's development through using the In-Motion app	1.6%	7.9%	28.6%	22.2%	39.7%
Using the In-Motion app made me more attentive to my child's development	7.9%	36.5%	46.0%	4.8%	4.8%

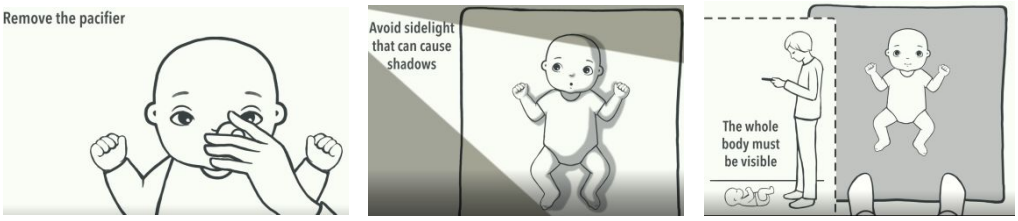

Figure 1: Screenshot of In-Motion instructional video showing examples of information about infant state, lighting and positioning of baby and person filming the baby.

Figure 2 Flow chart of infant/families with reasons for non-upload of videos.

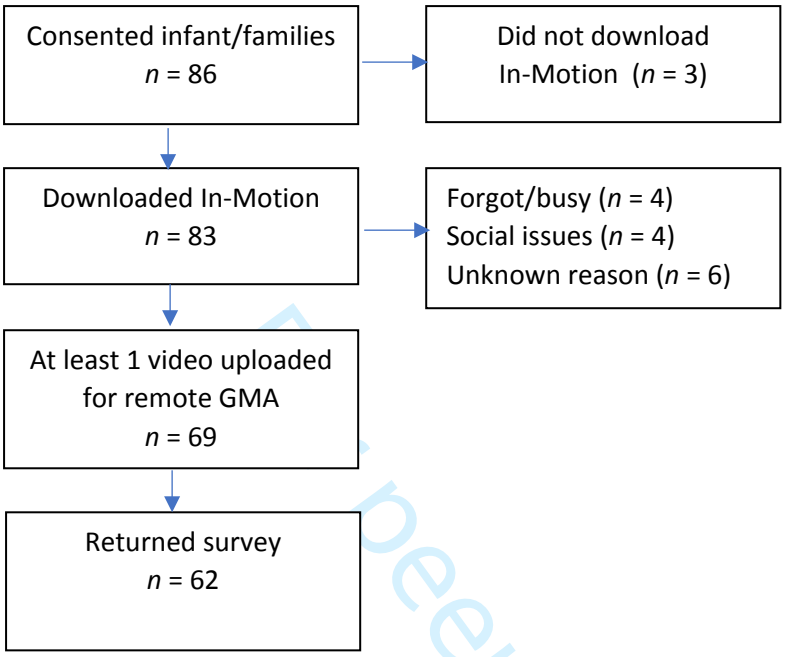

Figure 3 Accuracy of 7 estimated body points compared with manually annotated images

	%
Mean	80.9
Nose	93.3
Thorax	93.9
Right wrist	74.4
Left wrist	83.1
Pelvis	63
Right ankle	79.5
Left ankle	79.3

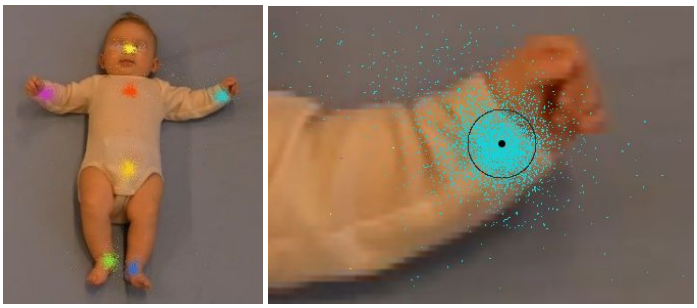

Figure 3: From left: Table with proportions of correct detected body point; illustration of the computer-based detections according to 7 body points; the distribution of the left wrist body point detections (blue dots) relative to the manually annotated landmark (black dot) where 10% of the infant head size is used as a threshold (black circle).

Appendix S1 – Parent survey

Thank you for taking the time to answer these questions. It takes about 10-15 minutes to answer them. If you have questions, you can contact …, email ……, or mobile telephone …….

1. Are you the child's mother or father?

- Mother
- Father

2. What is your marital status?

- Single
- Married/cohabitant
- Separated/divorced
- Widow/widower

3. What is your age and what is the age of your spouse/partner?

- You:
- Your spouse/partner:

4. How many brothers and sisters live with the child sometimes or always?

5. What type of mobile phone do you have?

- iPhone (iOS/Apple)
- Android

6. To what extent do you agree or disagree with each of these statements?

The In-Motion-App was generally easy to use

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to enter the information needed in the In-Motion-App

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

The reminders about when the child should be filmed were helpful

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

The number of reminders about when the child should be filmed was suitable

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

There were no technical problems with uploading and sending the videos

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to understand how I should stand and hold the telephone during the filming

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to keep the telephone still while I was filming

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to do the filming without disturbing the child

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to understand how my child should be dressed when filmed

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to understand how my child should be positioned and how the mat should be when I was going to film

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to follow the instructions about how the lighting should be during the filming

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

Filming my child for 3 minutes went smoothly

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

I felt safe about uploading video of my child

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

I become more worried about my child's development through using the In-Motion-App

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

Using the In-Motion app made me more attentive to my child's development

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

STROBE 2007 (v4) Statement—Checklist of items that should be included in reports of *cohort studies*

Section/Topic	Item #	Recommendation	Reported on page #
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract	1
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found	2
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	3
Objectives	3	State specific objectives, including any prespecified hypotheses	3
Methods			
Study design	4	Present key elements of study design early in the paper	3
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	4
Participants	6	(*a*) Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up	4
		(*b*) For matched studies, give matching criteria and number of exposed and unexposed	na
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	4-5
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	4-5
Bias	9	Describe any efforts to address potential sources of bias	5
Study size	10	Explain how the study size was arrived at	4
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	6
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding	6
		(*b*) Describe any methods used to examine subgroups and interactions	na
		(*c*) Explain how missing data were addressed	na
		(*d*) If applicable, explain how loss to follow-up was addressed	na
		(*e*) Describe any sensitivity analyses	na
Results			

Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	6
		(b) Give reasons for non-participation at each stage	6
		(c) Consider use of a flow diagram	6
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	7
		(b) Indicate number of participants with missing data for each variable of interest	na
		(c) Summarise follow-up time (eg, average and total amount)	na
Outcome data	15*	Report numbers of outcome events or summary measures over time	6-7
Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	na
		(*b*) Report category boundaries when continuous variables were categorized	na
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	na
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	6-7
Discussion			
Key results	18	Summarise key results with reference to study objectives	7
Limitations			
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	7
Generalisability	21	Discuss the generalisability (external validity) of the study results	7-8
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	8

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at www.strobe-statement.org.

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Title:

The In-Motion-App for remote General Movement Assessment: A multi-site observational study.

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Abstract

Objectives To determine whether videos taken by parents of their infants' spontaneous movements were in accordance with required standards in the In-Motion-App, and whether the videos could be remotely scored by a trained General Movement Assessment (GMA) observer. Additionally, to assess the feasibility of using home-based video recordings for automated tracking of spontaneous movements, and to examine parents' perceptions and experiences of taking videos in their homes.
Design The study was a multi-center prospective observational study.
Setting Parents/families of high-risk infants in tertiary care follow-up programs in Norway, Denmark and Belgium.
Methods Parents/families were asked to video record their baby in accordance with the In-Motion standards which were based on published GMA criteria and criteria covering lighting and stability of smartphone. Videos were evaluated as GMA "scorable" or "non-scorable" based on pre-defined criteria. The accuracy of a 7-point body tracker software was compared to manually annotated body key points. Parents were surveyed about the In-Motion-App information and clarity.
Participants The sample comprised 86 parents/families of high-risk infants.
Results The 86 parent/families returned 130 videos, and 121 (96%) of them were in accordance with the requirements for GMA assessment. The 7-point body tracker software detected more than 80% of body key point positions correctly. Most families found the instructions for filming their baby easy to follow, and more than 90% reported that they did not become more worried about their child's development through using the instructions.
Conclusions This study reveals that a short instructional video enabled parents to video record their infant's spontaneous movements in compliance with the standards required for remote GMA. Further, an accurate automated body point software detecting infant body landmarks in smartphone videos will facilitate clinical and research use soon. Home-based video recordings could be performed without worrying parents about their child's development.

Strengths and limitations of this study

- A cohort of families of high-risk infants frequently seen in NICU follow-up settings.
- In-Motion-App standards for remote General Movement Assessment (GMA) communicated through a simple and short animated video.
- Data from a motion tracking software on smartphone videos pioneering automatic and markerless infant motion capture.
- Study did not assess socio-demographic factors as reasons for families not to record or return videos.
- Study did not evaluate how markerless infant motion capture on smartphone videos can be used for prediction of CP outcome.

1. INTRODUCTION

Cerebral Palsy (CP) is the most common physical disability in childhood. Diagnosis is typically set between 12- and 24-months corrected age (1-3). Early developmental screening of high-risk infants to predict future neurological impairments is today a priority for clinicians and researchers, and most parents express interest in such neurodevelopmental screening (3, 4).

The General Movement Assessment (GMA) has been recommended in combination with magnetic resonance imaging (MRI) to achieve a CP diagnosis before 6 months corrected age in infants with newborn-detectable risk factors (3). Early detection of CP has the potential to improve the organization and resources used in follow-up screening at hospitals and reduce medical complications for children with CP. The fidgety type of general movements (GMs) observed at 9-20 weeks' corrected age has shown the highest predictive validity for later CP, compared to the writhing type of general movements observed before 9 weeks corrected age (5-8). Video recordings for GMA must follow requirements for infant state, position and clothing and are scored by certified and trained assessors (9). Such trained GMA observers also have the expertise needed to ensure that video recordings fulfill the requirements for a valid GMA (9).

Access to trained observers using the GMA in hospital-based follow-up programs is limited by geographical constraints and lack of GMA expertise (10). As health care and parents move into the digital age using smartphones to share videos via internet, opportunities to perform remote GMA have developed. Recently, the Baby Moves smartphone app was presented for remote GMA within research settings (10, 11). However, smartphone apps for health data capturing in clinical settings are rarely assessed and usability tested (12). To be feasible in a clinical follow-up setting, home-based video recordings must fulfill basic GMA requirements without the need for comprehensive parental training or guiding.

Video recordings by hand-held smartphones introduce movement artifacts in the camera. Computer-based methods for objective detection of infant GMs (13, 14) may be jeopardized by such artifacts. Our research group has recently presented a machine-learning model which predicted CP with high accuracy (sensitivity of 92%, specificity of 81%) performed by clinician using a stationary camera, comparable to observational GMA (15). Important shortcomings of the method were the need for manual and time-consuming body point annotations, as well as the need for a stationary camera. Hence, an automated 7-point body tracker has been developed by our group and needs to be validated on recordings taken with hand-held cameras.

Provided that GMs can be assessed, and computer-based infant body point tracking can be performed on videos taken with a hand-held camera, it is possible to perform remote GMA as well as automated infant body point tracking for a computer-based model for the prediction of CP. Thus, the In-Motion instructional video has been developed so that parents can perform home-based videos with quality standards feasible for remote GMA and automated infant body point tracking. Feasibility of the In-Motion instructional video was assessed in a multi-site study including families of high-risk infants from Norway, Denmark and Belgium.

The main aims of the study are as follows: 1) To determine whether videos taken by parents with a hand-held camera were in accordance with the standards set in the instructional video, and whether the videos could be scored by a trained GMA observer. 2) To assess the accuracy of a 7-point body tracker software based on the same recordings. 3) To describe parents' perceptions of the instructional video and filming their baby in a home environment.

2. METHODS

Design

Multi-center prospective observational study.

Patient and public involvement

The study protocol including the parental survey content was developed and designed collaboratively with representatives from The Norwegian Cerebral Palsy organization and The Norwegian Premature Association.

Participants

Parents of infants admitted to one of five participating level III-IV Neonatal Infant Care Units (NICU) in Norway (three hospitals including 13, 11 and 4 families, respectively), Denmark (one hospital including 43 families) and Belgium (one hospital including 15 families) from 2018-2019 (12 months recruitment period) were consecutively recruited at referral to the hospital follow-up program before discharge from the NICU. Families were recruited based on willingness to participate and the infant being evaluated as at high-risk of CP. In Norway and Denmark inclusion criteria were (a) Birth weight (BW) ≤1000 g (extremely low birth weight (ELBW) and/or gestational age (GA) <28 (extremely low GA (ELGA)), (b) neonatal arterial ischemic stroke, (c) neonatal encephalopathy, (d) other significant risk factors. In Belgium, only infants with GA <32 weeks or with perinatal stroke were included.

Data collection procedure

Included participants were assisted by a research physiotherapist/pediatrician at the time of inclusion to download and install the In-Motion-App by smartphone, containing the instructional video from Google Play or iTunes. They could ask any questions about the app and how to manage the software. They got information about the time window for performing two separate video recordings for their infant between 12^{+1} - 13^{+6} and 14^{+1} - 17^{+6} weeks post term age (PTA). The time points were defined to ensure GM videos from the fidgety movement's period. If no videos were returned from the families before 17^{+6} weeks PTA, the local study coordinator contacted the family by phone to ask the reason why they had not uploaded any videos. The app was linked to a secure online server hosted at St. Olavs Hospital in Trondheim, Norway, and was available for i-operating system (iOS) and Android. After the end of the second time window, the families were contacted by email with a link to an online Norwegian University of Science and Technology survey to collect information about their opinions using the app and the In-Motion instructional video.

The In-Motion-App and instructional video

The In-Motion-App and instructional video was designed by GMA trained personnel (LA, TF, RS, SO) at St. Olavs Hospital in Trondheim, Norway, for parents to give basic insight into recording standards needed for GMA and lighting and stability of camera. It was made as a short animation with simple drawn sequences containing a minimum of text. The instructional video was deployed to the parents by downloading the In-Motion-App developed for this study, and videos could be uploaded to be remotely assessed by a trained GMA observer.

After downloading the app and getting basic information from the local study coordinator, parents logged into the app with a username and a password. They typed in the first name of their child and the expected date of delivery (due date). The In-Motion-App generated two separate time windows between 12^{+1} - 13^{+6} and 14^{+1} - 17^{+6} weeks PTA and visualized them in a graphical timeline to show when videos should be taken. A red dot illustrated todays date placed on the timeline, helping parents to plan when to perform the video recordings. In addition, a pop-up message reminded parents to prepare for videos a week before the beginning of each time window. The In-Motion-App

was constructed in such a way that the video recording automatically stopped after 3 minutes and asked the parents whether to upload the video or not.

The instructional video was 2 minutes and 47 seconds long. Before taking videos, parents were told to look through the In-Motion instructional video which was available from the app menu. They could watch the video as many times as they wanted until they felt confident performing the recording. The main themes aimed at ensuring quality standards for remote GMA included: a) clothing of infant (just a diaper or a onesie), b) surface/underlay for infant (single-color blanket or rug), c) lighting (enough light avoiding sidelight that can cause shadows), d) state of infant (awake, alert, content, not disturbing baby, no pacifier), e) positioning (baby on floor- stand next to the baby's feet, whole body must be visible) and f) length of video (3 minutes). In addition, instructions were provided about how to keep the smartphone steady and ensure that the whole infant body was observable in the video image. Examples of some of the In-Motion instructional video themes are shown in Figure 1. Parents were asked to consecutively upload videos to the server at St. Olavs Hospital in Norway.

Assessment of video quality for remote GMA

Videos were assessed by a certified GMA observer with respect to the following standards (9): *1) GMA standards:* active movements (not hypokinetic), supine position, correct state, adequate clothing (diaper or a onesie), no disturbances during recording. *2) Additional In-Motion standards:* adequate light, whole body visible, feet of parent visible in video (ensuring correct position of smartphone camera, see picture to the right in Figure 1) and camera stability. Based on these standards, a classification was made by the same certified GMA observer as either "GMA scorable" if all standard criteria were fulfilled or "GMA non-scorable" if one or more standard criteria were inadequate. In addition, all videos were observed by the same GMA expert who also categorized yes/no whether the hand-held video had optimal stability, events of abrupt displacement, was predominantly unstable, whether an adequate underlay was used (firm, comfortable, large enough) and whether overall video image quality was sufficient (blurred/very blurred).

Assessment of General Movements

All videos classified as "GMA scorable" were consecutively assessed by one certified and experienced GMA observer that had passed advanced GMs courses under the General Movement Trust (LA). The use of one observer was chosen due to the study design not focusing on GMA and prediction of outcome. The observer had no knowledge about the infant's clinical history. According to Prechtl's method of assessment of general movements (9), fidgety movements were classified as continuous (FM++), intermittent (FM+), sporadic (FM+/-), abnormal (Fa) or absent (FM-).

In-Motion body point tracking

A subset of 66 videos from 36 infants recorded by the In-Motion-App by September 19th 2018 was used for automatic infant motion tracking to detect the displacement of 7-body points (nose, thorax (center between shoulders), wrists, pelvis, and ankles). Eighty and 20% out of 5493 frames from these videos were selected by random and chosen due to body part occlusions for motion tracking testing, respectively. The infant motion tracker algorithm consists of a convolutional neural net trained on 7-body points on 14900 video frames on high-risk infants that had participated in another study from our group (8). For further technical details of the trained convolutional neural net, the reader is referred to Groos et al (16). All 7 body points in the 5493 video test frames used in this study were manually annotated and used as ground truth, comparing them with the automatic body point positions for evaluation. The performance of the infant motion tracker is reported as percentage of points within a circular area around the manually annotated body point for the 5493 test frames. In accordance with the established metric for evaluating pose-estimation (17), size of

the circular area was set to 10% of the infant head size and was normalized to adjust for different scaling (i.e., video zoom) (Figure 2).

Survey

Parents' opinions about the In-Motion-App and instructional video were collected using SelectSurvey v.4 software (ClassApps Inc., www.classapps.com) in Norway, operated by Norwegian University of Science and Technology (NTNU). In Denmark, parents' opinions were collected using Smart Trails data management software (MEDEI ApS. www.medei.dk) and in Belgium the survey was collected by post. The survey was sent to the families by a link in an email within one week after the last video was returned. The survey questions were customised for the In-Motion-App based on a tool developed by Jin and Kim in 2015 (18) and a survey used in a similar study on the Baby Moves App (11). It contained questions based on forced-choice questions covering the themes; 1) In-Motion-App, 2) In-Motion standards for remote GMA and 3) parental worries. A statement was made, and the parents indicated agreement or disagreement with the statement on a 5-point scale (Appendix S1, supplementary information).

Data analysis

Data were analyzed using SPSS statistics version 26.0 (IBM SPSS Statistics, Chicago, IL). The data are presented as numbers with proportion (%) or mean with standard deviation (SD) and range. Differences between infants with returned and no returned videos were analyzed using the Mann-Whitney U Test for continuous variables and Chi-square test and Fisher's Exact test for dichotomous variables. The accuracy of the 7-point body tracker was presented as the distribution of body point detections relative to the manually annotated body points, where 10% of the infant head size was used as a threshold.

3. RESULTS

In total, 86 infants/families were recruited and 17 (19.8%) out of them did not submit any video (Figure 3), leaving 69 families with 130 videos for analysis.

Twenty-eight (32.6%) families were included from three different hospitals in Norway, 43 (50%) from one hospital in Copenhagen, Denmark and 15 (17.4%) from one hospital in Gent, Belgium. Infant/family characteristics are shown in table 1.

Table 1 Summary of infant/family characteristics

	Total N (%) (*n*=86)	N (%) video responders (*n*=69)	N (%) no video responders (*n*=17)	*p*-value
Demographics				
Boys, n (%)	51(59.3)	42(60.9)	9(52.9)	0.32
BW, mean (SD), grams	1952(1107)	1915(1124)	2105(1055)	0.67
GA, mean (SD), weeks	32.4(5.3)	32.3(5.4)	33.4(4.7)	0.43
Risk group				
Birth weight (BW) ≤1000 g and/or gestational age (GA) <28	25(29.1)	20(29)	5(29.4)	0.77
Neonatal arterial ischemic stroke	11(12.8)	4(5.8)	7(41.2)	0.001
Hypoxic ischemic encephalopathy	20(23.3)	17(24.6)	3(17.6)	0.06
Others	32(37.2)	27(39.1)	5(29.4)	0.38
		Infant families (*n*=63)		
Socio-demographic data*				
Mother relation (survey), n (%)		48(76.2)		
Married/cohabitant family, n (%)		59(93.7)		
Age mother/farther, mean (SD, range)		31.8(5.5,21-6)		
Age farther, mean (SD, range)		33.9 (6.9, 22-59)		
Single child, n (%)		32 (51.6)		
iOS vs. Android				
iOS users, n (%)		41(65.1)		

3.1 Video recordings

Two (1.5%) out of 130 videos were shorter than 3 minutes (1 minute and 12 seconds and 2 minutes and 17 seconds). The mean PTA at video recording was 14.5 weeks (SD 2.28, range 8.1-23.6 weeks).

Fifteen (11.7%) families returned one recording, 49 (71%) two recordings, 4 (5.8%) three recordings, and 1 (1.4%) six recordings. One-hundred and seventeen (90%) videos were returned within the expected time window between 12^{+1} - 17^{+6} weeks PTA. Two (1.5%) families returned 2 and 3 videos, respectively, which were all taken outside the time window (week 8, 10, and 21 and 23, respectively). Eleven families (8.5%) with videos from within the requested time window, had additional videos taken outside the expected time windows. Six videos from one family were taken at two different days; four videos in weeks 12 and two videos in week 14 PTA.

3.2 Remote General Movement Assessment

Among the six videos returned from one family, the first one from each of the two days was selected for GMA analysis. Exclusion of the remaining 4 videos and additional 4 videos excluded due to PTA

outside of the age required for assessment of FMs gave a total of 122 videos available for quality assessment.

One hundred and twenty-one (99%) out of 122 videos which were returned within the required time window were classified as GMA scorable. The video that was non-scorable had infant in side lying position. Details about compliance to the In-Motion standards are shown in Table 2.

Table 2 Compliance to In-Motion standards (n= 126)

	Active movements (not hypokinetic)	Supine position	Correct state	No disturbances during recording	Adequate clothing	Adequate light	Whole body visible	Feet of parents visible
N (%)	126 (100)	125 (99.2)	122 (96.8)	124 (98.4)	125 (99.2)	124 (98.4)	124 (98.4)	116 (92.1)*
		Optimal stability	Abrupt displacement	Predominantly unstable	Correct base of support		Image quality	
						Clear	Blurred	Very blurred
N (%)		80 (63.5)	26 (20.6)	22 (17.5)	119 (94.4)	114 (90.5)	11 (8.7)	1 (0.8)

*3 missing data.

3.3 General Movement Assessment

Of the 121 videos classified as GMA scorable, 3 (2.4%) videos were classified with exaggerated (Fa), 3 (2.4%) with absent (FM-), and 7 (5.6%) with sporadic (FM+/-) FMs. Eighty-seven (69%) and 21 (16.7%) videos were classified with intermittent (FM+) and continuous (FM++) FMs, respectively.

3.4 Computer-based body point tracking

The proportion of correctly predicted left wrist key point from 5493 tested video images was 83.15%. Details of accuracy of 7 predicted body points and mean value for all points are shown in Figure 2.

3.5 Parent responses

Survey responses were received from 64 (92.8%) families of the 69 families who returned at least one video. Fifty-four (84.3%) of them observed the instructional video one or two times before filming their baby. No families returned a video without training on filming their baby first.

The majority of the survey respondents found the In-Motion-App easy to use. All respondents agreed or strongly agreed that it was easy to understand how to stand and hold the smartphone during the filming. Details about family responses are shown in Table 3. Fifty-seven (90.5%) families strongly disagreed, disagreed or neither disagreed nor agreed that they did become more worried about their child's development through using the In-Motion instructions.

Table 3 Parents' responses to the In-Motion-App and instructional video

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree
In-Motion_App					
The In-Motion app was generally easy to use	58.7%	33.3%	3.2%	4.8%	0.0%
It was easy to enter the information needed in the In-Motion app	46.0%	41.3%	11.1%	1.6%	0.0%
The reminders about when the child should be filmed were helpful	44.4%	41.3%	6.3%	6.3%	1.6%
The number of reminders about when the child should be filmed was suitable	25.4%	39.7%	15.9%	14.3%	4.8%
There were no technical problems with uploading and sending the videos	55.6%	22.2%	6.3%	4.8%	11.1%
In-Motion standards for remote GMA					
It was easy to understand how I should stand and hold the telephone during the filming	60.0%	40.0%	0.0%	0.0%	0.0%
It was easy to keep the telephone still while I was filming	16.7%	46.7%	30.0%	6.7%	0.0%
It was easy to do the filming without disturbing the child	36.5%	42.9%	9.5%	11.1%	0.0%
It was easy to understand how my child should be dressed when filmed	65.1%	33.3%	0.0%	1.6%	0.0%
It was easy to understand how my child should be positioned and how the mat should be when I was going to film	63.5%	28.6%	4.8%	3.2%	0.0%
It was easy to follow the instructions about how the lighting should be during the filming	36.5%	38.1%	14.3%	11.1%	0.0%
Filming my child for 3 minutes went smoothly	39.7%	39.7%	19.0%	1.6%	0.0%
Parental worries					
I felt safe about uploading video of my child	63.5%	27.0%	7.9%	1.6%	0.0%
I became more worried about my child's development through using the In-Motion app	1.6%	7.9%	28.6%	22.2%	39.7%
Using the In-Motion app made me more attentive to my child's development	7.9%	36.5%	46.0%	4.8%	4.8%

DISCUSSION

In this study, more than 95% of families with high-risk infants filming their baby at home, returned at least one video that was in accordance with the In-Motion standards for remote GMA. Most families found the In-Motion-App easy to use and the instructions for filming easy to follow, and less than 10% of respondents became worried through using the In-Motion-App. Despite the use of hand-held smartphones introducing movement artifacts in the video image, our computer-based 7-point body tracker detected positions of the body points with high accuracy. To the best of our knowledge, this is the first automatic infant body point tracker that is tested on video recordings from hand-held smartphones.

This study has several strengths. First, it included families of high-risk infants frequently seen in NICU follow-up settings. We argue that this makes our findings robust and generalizable to comparable clinical settings. Second, the communication of In-Motion standards through a simple and short animated video, makes the instructions easily applicable to a broad range of different clinical settings. Furthermore, the accuracy of the 7-point body tracker software with the use of smartphone videos makes it pioneering in the field of automatic and markerless infant motion capture compared to other studies (19, 20). This facilitates further development of methods for early automated detection of CP based on smartphone videos. Finally, the design of the study using an experienced and certified GMA observer for evaluation of video quality and a survey with very high response rate, makes the study quality high and the results trustworthy.

There are also several limitations. First, almost 20% of the included families did not return any video. This study was not designed to evaluate reasons for not recording or returning videos. We can, therefore, only conclude on the quality of returned videos. The questions in our survey may also have limitations, mainly covering topics favorable to participants returning videos, participating in follow-up and smartphone usage, giving little or reduced information about responders with low mobile health technology usage. Hence, problems encountered by families who did not record or return any video need to be further explored. Our findings are in accordance with the study by Kwong et al (11), where 24% of families did not return any video using the Baby Moves App. These findings indicate that home-based video recordings for remote GMA is not a solution for all families and that it might be difficult in a clinical setting to know beforehand which families will return a video or not. Furthermore, 13 (19%) families returned one or several videos outside the required time window needed for a valid GMA. More than 90% of respondents found the reminders in the App helpful, but one fifth disagreed that the number of reminders were appropriate. These findings indicate a limitation in the design of the app reminders and lack of programmed filming windows parameters. These app functionalities need to be improved in a process involving the users.

Second, our study comprised five different hospital sites in three different countries, and information provided to families when downloading the App may have differed. Additionally, there is a risk that the research personnel could have given more information and assistance to families than will be common in an ordinary clinical setting. Our study setting could therefore be in slight contrast to an ordinary clinical setting, where adapted and flexible family information is needed due to challenges in reduced participation in neurodevelopmental follow-up (21) and racial and socioeconomic differences in mobile health technology usage (22).

Third, even though almost 20% of the hand-held videos in our study were classified as predominantly unstable, the GMA expert considered the videos to be GMA scorable. There is a risk that more videos might have been classified as non-scorable by another GMA observer with less training and experience or if there had been several GMA experts observing the same videos. To the best of our knowledge, there is only one study protocol planning to assess the predictive validity of

GMA from videos taken with hand-held smartphones for CP outcome (10). Hence, further studies on the use of smartphones for GMA are needed.

Finally, our computer-based 7-point body tracker showed accurate estimations compared to manual annotations on the video image. However, further studies must explore how selection of body points, tracked body point accuracy and movement artifacts in camera will influence a machine-learning model for prediction of CP from smartphone video recordings.

This study facilitates and contributes to the use of smartphone technology for video recordings and remote GMA. Consequently, it will contribute to giving high-risk infants and their families equal access to GMA as an accurate method for early identification of CP, without geographical constraints. The use of early remote medical assessment will improve the organization and resources used in follow-up screening at hospitals and have the potential to reduce medical complications for children with CP due to early detection. A clinical feasible computer-based movement analysis with equal accuracy as GMA, will greatly reduce the need for specialized GMA observers and provide an innovative resource-effective diagnostic measure.

Acknowledgements
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Contributors
Conceptualization; LA, RS, HP, OMS, TF, SO, WS, BE, CB, Data curation; LA, RS, EI, DG, Formal analysis; LA, RS, EI, DG, Funding acquisition; LA, RS, Methodology; LA, RS, EI, DG, TF, SO, Project administration; LA, BE, HP, AP, AB, CB, RS, KC, Resources; LA, TF, SO, AB, AP, HP, OMS, BE, WS, KC, Writing-original draft; LA, RS, EI, DG, Writing-review & editing; LA, RS, EI, DG, TF, SO, BE, WS, AB, AP, CB, HP, OMS, KC.

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Competing interests
None declared.

Patient consent
Obtained in all participants.

Ethics approval
All infant/families provided written informed consent and ethics was approved by the regional committee for medical and health research ethics (REC Central-Committee 2017/913) in Norway. The study sites in Denmark and Belgium also had approvals from their local Institutional Review boards. Study registration in ClinicalTrials.gov Protocol Record 2017/913.

Provenance and peer review
Not commissioned; external peer reviewed.

Data availability statement

No data are available. Data from this study are not available for sharing due to ethical approval requirements. Researchers interested in collaboration should contact the corresponding author with their expression of interest.

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Figure title – Figure 1

Figure 1: Screenshot of In-Motion instructional video showing examples of information about infant state, lighting and positioning of baby and person filming the baby.

Figure title and legend – Figure 2

Figure 2 Accuracy of 7 estimated body points compared with manually annotated images

Figure 2: From left: Table with proportions of correct detected body point; illustration of the computer-based detections according to 7 body points; the distribution of the left wrist body point detections (blue dots) relative to the manually annotated landmark (black dot) where 10% of the infant head size is used as a threshold (black circle).

Figure title – Figure 3

Figure 3 Flow chart of infant/families with reasons for non-upload of videos.

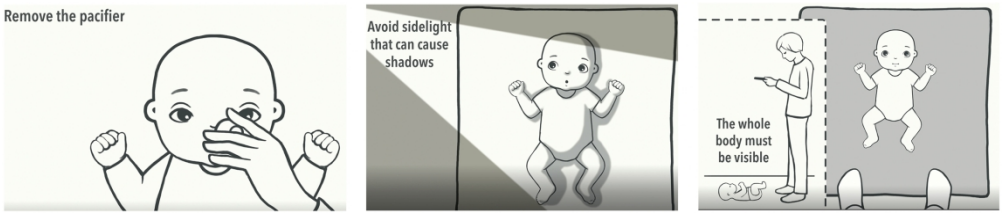

Figure 1: Screenshot of In-Motion instructional video showing examples of information about infant state, lighting and positioning of baby and person filming the baby.

209x44mm (300 x 300 DPI)

	%
Mean	80.9
Nose	93.3
Thorax	93.9
Right wrist	74.4
Left wrist	83.1
Pelvis	63
Right ankle	79.5
Left ankle	79.3

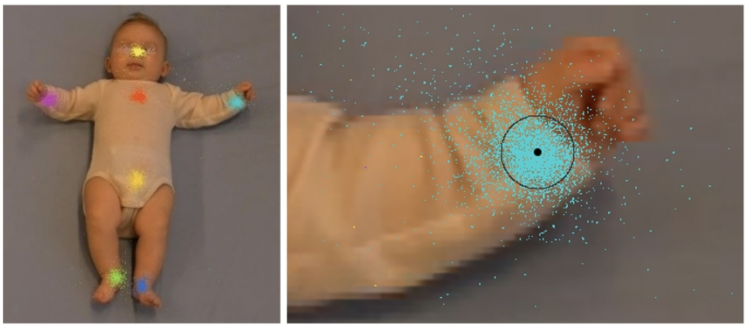

Figure 2 Accuracy of 7 estimated body points compared with manually annotated images
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209x68mm (300 x 300 DPI)

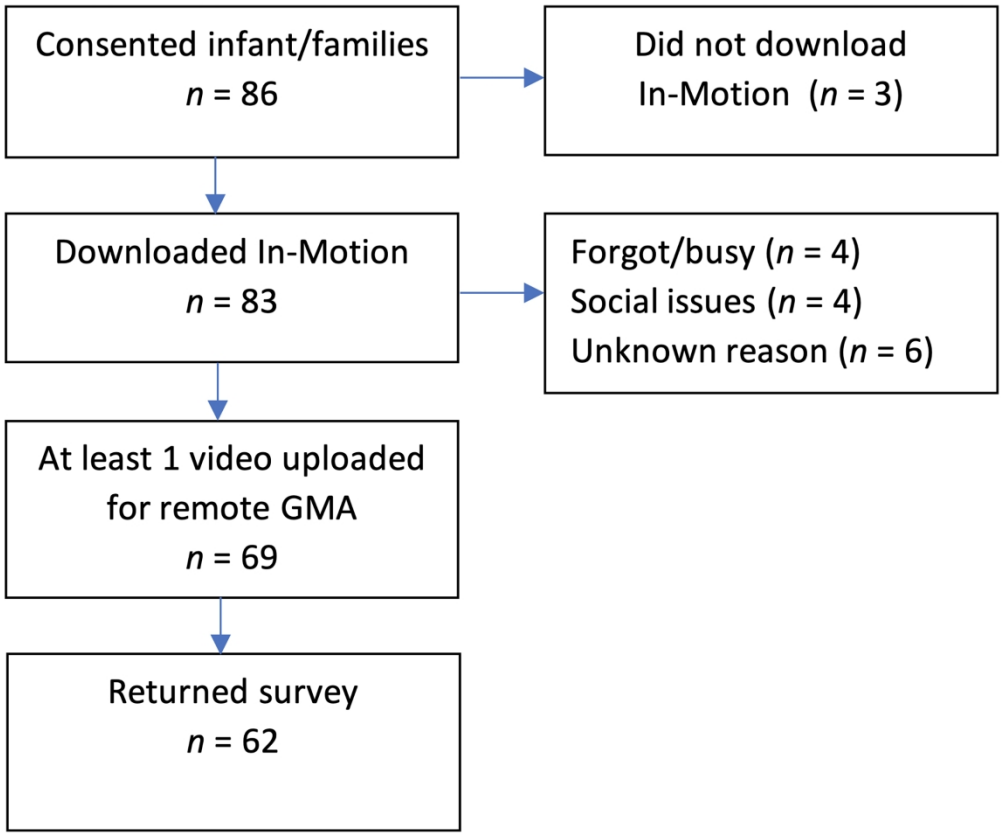

Figure 3 Flow chart of infant/families with reasons for non-upload of videos.

209x174mm (300 x 300 DPI)

Appendix S1 – Parent survey

Thank you for taking the time to answer these questions. It takes about 10-15 minutes to answer them. If you have questions, you can contact …, email ……, or mobile telephone …….

1. Are you the child's mother or father?

- Mother
- Father

2. What is your marital status?

- Single
- Married/cohabitant
- Separated/divorced
- Widow/widower

3. What is your age and what is the age of your spouse/partner?

- You:
- Your spouse/partner:

4. How many brothers and sisters live with the child sometimes or always?

5. What type of mobile phone do you have?

- iPhone (iOS/Apple)
- Android

6. To what extent do you agree or disagree with each of these statements?

The In-Motion-App was generally easy to use

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to enter the information needed in the In-Motion-App

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

The reminders about when the child should be filmed were helpful

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

The number of reminders about when the child should be filmed was suitable

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

There were no technical problems with uploading and sending the videos

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to understand how I should stand and hold the telephone during the filming

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to keep the telephone still while I was filming

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to do the filming without disturbing the child

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to understand how my child should be dressed when filmed

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to understand how my child should be positioned and how the mat should be when I was going to film

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to follow the instructions about how the lighting should be during the filming

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

Filming my child for 3 minutes went smoothly

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

I felt safe about uploading video of my child

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

I become more worried about my child's development through using the In-Motion-App

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

Using the In-Motion app made me more attentive to my child's development

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

STROBE 2007 (v4) Statement—Checklist of items that should be included in reports of *cohort studies*

Section/Topic	Item #	Recommendation	Reported on page #
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract	1
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found	2
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	3
Objectives	3	State specific objectives, including any prespecified hypotheses	3
Methods			
Study design	4	Present key elements of study design early in the paper	3
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	4
Participants	6	(*a*) Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up	4
		(*b*) For matched studies, give matching criteria and number of exposed and unexposed	na
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	4-5
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	4-5
Bias	9	Describe any efforts to address potential sources of bias	5
Study size	10	Explain how the study size was arrived at	4
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	6
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding	6
		(*b*) Describe any methods used to examine subgroups and interactions	na
		(*c*) Explain how missing data were addressed	na
		(*d*) If applicable, explain how loss to follow-up was addressed	na
		(*e*) Describe any sensitivity analyses	na
Results			

Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	6
		(b) Give reasons for non-participation at each stage	6
		(c) Consider use of a flow diagram	6
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	7
		(b) Indicate number of participants with missing data for each variable of interest	na
		(c) Summarise follow-up time (eg, average and total amount)	na
Outcome data	15*	Report numbers of outcome events or summary measures over time	6-7
Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	na
		(*b*) Report category boundaries when continuous variables were categorized	na
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	na
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	6-7
Discussion			
Key results	18	Summarise key results with reference to study objectives	7
Limitations			
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	7
Generalisability	21	Discuss the generalisability (external validity) of the study results	7-8
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	8

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at www.strobe-statement.org.

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The In-Motion-App for remote General Movement Assessment: A multi-site observational study.

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Title:

The In-Motion-App for remote General Movement Assessment: A multi-site observational study.

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Abstract

Objectives To determine whether videos taken by parents of their infants' spontaneous movements were in accordance with required standards in the In-Motion-App, and whether the videos could be remotely scored by a trained General Movement Assessment (GMA) observer. Additionally, to assess the feasibility of using home-based video recordings for automated tracking of spontaneous movements, and to examine parents' perceptions and experiences of taking videos in their homes.
Design The study was a multi-center prospective observational study.
Setting Parents/families of high-risk infants in tertiary care follow-up programs in Norway, Denmark and Belgium.
Methods Parents/families were asked to video record their baby in accordance with the In-Motion standards which were based on published GMA criteria and criteria covering lighting and stability of smartphone. Videos were evaluated as GMA "scorable" or "non-scorable" based on pre-defined criteria. The accuracy of a 7-point body tracker software was compared to manually annotated body key points. Parents were surveyed about the In-Motion-App information and clarity.
Participants The sample comprised 86 parents/families of high-risk infants.
Results The 86 parent/families returned 130 videos, and 121 (96%) of them were in accordance with the requirements for GMA assessment. The 7-point body tracker software detected more than 80% of body key point positions correctly. Most families found the instructions for filming their baby easy to follow, and more than 90% reported that they did not become more worried about their child's development through using the instructions.
Conclusions This study reveals that a short instructional video enabled parents to video record their infant's spontaneous movements in compliance with the standards required for remote GMA. Further, an accurate automated body point software detecting infant body landmarks in smartphone videos will facilitate clinical and research use soon. Home-based video recordings could be performed without worrying parents about their child's development.

Strengths and limitations of this study

- A cohort of families of high-risk infants frequently seen in NICU follow-up settings.
- In-Motion-App standards for remote General Movement Assessment (GMA) communicated through a simple and short animated video.
- Data from a motion tracking software on smartphone videos pioneering automatic and markerless infant motion capture.
- Study did not assess socio-demographic factors as reasons for families not to record or return videos.
- Study did not evaluate how markerless infant motion capture on smartphone videos can be used for prediction of CP outcome.

1. INTRODUCTION

Cerebral Palsy (CP) is the most common physical disability in childhood. Diagnosis is typically set between 12- and 24-months corrected age (1-3). Early developmental screening of high-risk infants to predict future neurological impairments is today a priority for clinicians and researchers, and most parents express interest in such neurodevelopmental screening (3, 4).

The General Movement Assessment (GMA) has been recommended in combination with magnetic resonance imaging (MRI) to achieve a CP diagnosis before 6 months corrected age in infants with newborn-detectable risk factors (3). Early detection of CP has the potential to improve the organization and resources used in follow-up screening at hospitals and reduce medical complications for children with CP. The fidgety type of general movements (GMs) observed at 9-20 weeks' corrected age has shown the highest predictive validity for later CP, compared to the writhing type of general movements observed before 9 weeks corrected age (5-8). Video recordings for GMA must follow requirements for infant state, position and clothing and are scored by certified and trained assessors (9). Such trained GMA observers also have the expertise needed to ensure that video recordings fulfill the requirements for a valid GMA (9).

Access to trained observers using the GMA in hospital-based follow-up programs is limited by geographical constraints and lack of GMA expertise (10). As health care and parents move into the digital age using smartphones to share videos via internet, opportunities to perform remote GMA have developed. Recently, the Baby Moves smartphone app was presented for remote GMA within research settings (10, 11). However, smartphone apps for health data capturing in clinical settings are rarely assessed and usability tested (12). To be feasible in a clinical follow-up setting, home-based video recordings must fulfill basic GMA requirements without the need for comprehensive parental training or guiding.

Video recordings by hand-held smartphones introduce movement artifacts in the camera. Computer-based methods for objective detection of infant GMs (13, 14) may be jeopardized by such artifacts. Our research group has recently presented a machine-learning model which predicted CP with high accuracy (sensitivity of 92%, specificity of 81%) performed by clinician using a stationary camera, comparable to observational GMA (15). Important shortcomings of the method were the need for manual and time-consuming body point annotations, as well as the need for a stationary camera. Hence, an automated 7-point body tracker has been developed by our group and needs to be validated on recordings taken with hand-held cameras.

Provided that GMs can be assessed, and computer-based infant body point tracking can be performed on videos taken with a hand-held camera, it is possible to perform remote GMA as well as automated infant body point tracking for a computer-based model for the prediction of CP. Thus, the In-Motion instructional video has been developed so that parents can perform home-based videos with quality standards feasible for remote GMA and automated infant body point tracking. Feasibility of the In-Motion instructional video was assessed in a multi-site study including families of high-risk infants from Norway, Denmark and Belgium.

The main aims of the study are as follows: 1) To determine whether videos taken by parents with a hand-held camera were in accordance with the standards set in the instructional video, and whether the videos could be scored by a trained GMA observer. 2) To assess the accuracy of a 7-point body tracker software based on the same recordings. 3) To describe parents' perceptions of the instructional video and filming their baby in a home environment.

2. METHODS

Design

Multi-center prospective observational study.

Patient and public involvement

The study protocol including the parental survey content was developed and designed collaboratively with representatives from The Norwegian Cerebral Palsy organization and The Norwegian Premature Association.

Participants

Parents of infants admitted to one of five participating level III-IV Neonatal Infant Care Units (NICU) in Norway (three hospitals including 13, 11 and 4 families, respectively), Denmark (one hospital including 43 families) and Belgium (one hospital including 15 families) from 2018-2019 (12 months recruitment period) were consecutively recruited at referral to the hospital follow-up program before discharge from the NICU. Families were recruited based on willingness to participate and the infant being evaluated as at high-risk of CP. In Norway and Denmark inclusion criteria were (a) Birth weight (BW) ≤1000 g (extremely low birth weight (ELBW) and/or gestational age (GA) <28 (extremely low GA (ELGA)), (b) neonatal arterial ischemic stroke, (c) neonatal encephalopathy, (d) other significant risk factors. In Belgium, only infants with GA <32 weeks or with perinatal stroke were included.

Data collection procedure

Included participants were assisted by a research physiotherapist/pediatrician at the time of inclusion to download and install the In-Motion-App by smartphone, containing the instructional video from Google Play or iTunes. They could ask any questions about the app and how to manage the software. They got information about the time window for performing two separate video recordings for their infant between 12^{+1} - 13^{+6} and 14^{+1} - 17^{+6} weeks post term age (PTA) (11). The time points were defined to ensure GM videos from the fidgety movement's period. If no videos were returned from the families before 17^{+6} weeks PTA, the local study coordinator contacted the family by phone to ask the reason why they had not uploaded any videos. The app was linked to a secure online server hosted at St. Olavs Hospital in Trondheim, Norway, and was available for i-operating system (iOS) and Android. After the end of the second time window, the families were contacted by email with a link to an online Norwegian University of Science and Technology survey to collect information about their opinions using the app and the In-Motion instructional video.

The In-Motion-App and instructional video

The In-Motion-App and instructional video was designed by GMA trained personnel (LA, TF, RS, SO) at St. Olavs Hospital in Trondheim, Norway, for parents to give basic insight into recording standards needed for GMA and lighting and stability of camera. It was made as a short animation with simple drawn sequences containing a minimum of text. The instructional video was deployed to the parents by downloading the In-Motion-App developed for this study, and videos could be uploaded to be remotely assessed by a trained GMA observer.

After downloading the app and getting basic information from the local study coordinator, parents logged into the app with a username and a password. They typed in the first name of their child and the expected date of delivery (due date). The In-Motion-App generated two separate time windows between 12^{+1} - 13^{+6} and 14^{+1} - 17^{+6} weeks PTA (11) and visualized them in a graphical timeline to show when videos should be taken. A red dot illustrated todays date placed on the timeline, helping parents to plan when to perform the video recordings. In addition, a pop-up message reminded parents to prepare for videos a week before the beginning of each time window (11). The In-Motion-

App was constructed in such a way that the video recording automatically stopped after 3 minutes and asked the parents whether to upload the video or not.

The instructional video was 2 minutes and 47 seconds long. Before taking videos, parents were told to look through the In-Motion instructional video which was available from the app menu. They could watch the video as many times as they wanted until they felt confident performing the recording. The main themes aimed at ensuring quality standards for remote GMA included: a) clothing of infant (just a diaper or a onesie), b) surface/underlay for infant (single-color blanket or rug), c) lighting (enough light avoiding sidelight that can cause shadows), d) state of infant (awake, alert, content, not disturbing baby, no pacifier), e) positioning (baby on floor- stand next to the baby's feet, whole body must be visible) and f) length of video (3 minutes). In addition, instructions were provided about how to keep the smartphone steady and ensure that the whole infant body was observable in the video image. Examples of some of the In-Motion instructional video themes are shown in Figure 1. Parents were asked to consecutively upload videos to the server at St. Olavs Hospital in Norway.

Assessment of video quality for remote GMA

Videos were assessed by a certified GMA observer with respect to the following standards (9): *1) GMA standards:* active movements (not hypokinetic), supine position, correct state, adequate clothing (diaper or a onesie), no disturbances during recording. *2) Additional In-Motion standards:* adequate light, whole body visible, feet of parent visible in video (ensuring correct position of smartphone camera, see picture to the right in Figure 1) and camera stability. Based on these standards, a classification was made by the same certified GMA observer as either "GMA scorable" if all standard criteria were fulfilled or "GMA non-scorable" if one or more standard criteria were inadequate. In addition, all videos were observed by the same GMA expert who also categorized yes/no whether the hand-held video had optimal stability, events of abrupt displacement, was predominantly unstable, whether an adequate underlay was used (firm, comfortable, large enough) and whether overall video image quality was sufficient (blurred/very blurred).

Assessment of General Movements

All videos classified as "GMA scorable" were consecutively assessed by one certified and experienced GMA observer that had passed advanced GMs courses under the General Movement Trust (LA). The use of one observer was chosen due to the study design not focusing on GMA and prediction of outcome. The observer had no knowledge about the infant's clinical history. According to Prechtl's method of assessment of general movements (9), fidgety movements were classified as continuous (FM++), intermittent (FM+), sporadic (FM+/-), abnormal (Fa) or absent (FM-).

In-Motion body point tracking

The infant motion tracker algorithm consists of a convolutional neural network trained on 7-body points on 14900 video frames on high-risk infants that had participated in another study from our group (8). For further technical details of the previous trained convolutional neural net, the reader is referred to Groos et al (16). To evaluate the infant motion tracker, 5493 video frames was selected from a subset of 66 videos from 36 infants, recorded by the In-Motion-App by September 19th 2018. Eighty percent out of the selected 5493 frames were selected by random. The other 20% were selected manually in order to include body part occlusions (for example right wrist occluded behind left wrist) that may be challenging to track. The performance of the infant motion tracker was assessed and reported by the following three steps: First, the automatic motion tracking was performed to detect the position of 7-body points (nose, thorax (center between shoulders), wrists, pelvis, and ankles) in each of the 5493 video frames. Secondly, all 7 body points in the 5493 selected video frames was manually annotated. These manually annotations are the ground truth for the

evaluation of the infant motion tracker. Thirdly, the performance of the infant motion tracker is reported as percentage of points within a circular area centered at the manually annotated body point for the 5493 frames. In accordance with the established metric for evaluating pose-estimation (17), radius of the circular area was set to 10% of the infant head size and was normalized to adjust for different scaling (i.e., video zoom) (Figure 2).

Survey

Parents' opinions about the In-Motion-App and instructional video were collected using SelectSurvey v.4 software (ClassApps Inc., www.classapps.com) in Norway, operated by Norwegian University of Science and Technology (NTNU). In Denmark, parents' opinions were collected using Smart Trails data management software (MEDEI ApS. www.medei.dk) and in Belgium the survey was collected by post. The survey was sent to the families by a link in an email within one week after the last video was returned. The survey questions were customised for the In-Motion-App based on a tool developed by Jin and Kim in 2015 (18) and a survey used in a similar study on the Baby Moves App (11). It contained questions based on forced-choice questions covering the themes; 1) In-Motion-App, 2) In-Motion standards for remote GMA and 3) parental worries. A statement was made, and the parents indicated agreement or disagreement with the statement on a 5-point scale (Appendix S1, supplementary information).

Data analysis

Data were analyzed using SPSS statistics version 26.0 (IBM SPSS Statistics, Chicago, IL). The data are presented as numbers with proportion (%) or mean with standard deviation (SD) and range. Differences between infants with returned and no returned videos were analyzed using the Mann-Whitney U Test for continuous variables and Chi-square test and Fisher's Exact test for dichotomous variables. The accuracy of the 7-point body tracker was presented as the distribution of body point detections relative to the manually annotated body points, where 10% of the infant head size was used as a threshold.

3. RESULTS

In total, 86 infants/families were recruited and 17 (19.8%) out of them did not submit any video (Figure 3), leaving 69 families with 130 videos for analysis.

Twenty-eight (32.6%) families were included from three different hospitals in Norway, 43 (50%) from one hospital in Copenhagen, Denmark and 15 (17.4%) from one hospital in Gent, Belgium. Infant/family characteristics are shown in table 1.

Table 1 Summary of infant/family characteristics

	Total N (%) (*n*=86)	N (%) video responders (*n*=69)	N (%) no video responders (*n*=17)	*p*-value
Demographics				
Boys, n (%)	51(59.3)	42(60.9)	9(52.9)	0.32
BW, mean (SD), grams	1952(1107)	1915(1124)	2105(1055)	0.67
GA, mean (SD), weeks	32.4(5.3)	32.3(5.4)	33.4(4.7)	0.43
Risk group				
Birth weight (BW) ≤1000 g and/or gestational age (GA) <28	25(29.1)	20(29)	5(29.4)	0.77
Neonatal arterial ischemic stroke	11(12.8)	4(5.8)	7(41.2)	0.001
Hypoxic ischemic encephalopathy	20(23.3)	17(24.6)	3(17.6)	0.06
Others	32(37.2)	27(39.1)	5(29.4)	0.38
		Infant families (*n*=63)		
Socio-demographic data*				
Mother relation (survey), n (%)		48(76.2)		
Married/cohabitant family, n (%)		59(93.7)		
Age mother/farther, mean (SD, range)		31.8(5.5,21-6)		
Age farther, mean (SD, range)		33.9 (6.9, 22-59)		
Single child, n (%)		32 (51.6)		
iOS vs. Android				
iOS users, n (%)		41(65.1)		

3.1 Video recordings

Two (1.5%) out of 130 videos were shorter than 3 minutes (1 minute and 12 seconds and 2 minutes and 17 seconds). The mean PTA at video recording was 14.5 weeks (SD 2.28, range 8.1-23.6 weeks).

Fifteen (11.7%) families returned one recording, 49 (71%) two recordings, 4 (5.8%) three recordings, and 1 (1.4%) six recordings. One-hundred and seventeen (90%) videos were returned within the expected time window between 12^{+1} - 17^{+6} weeks PTA. Two (1.5%) families returned 2 and 3 videos, respectively, which were all taken outside the time window (week 8, 10, and 21 and 23, respectively). Eleven families (8.5%) with videos from within the requested time window, had additional videos taken outside the expected time windows. Six videos from one family were taken at two different days; four videos in weeks 12 and two videos in week 14 PTA.

3.2 Remote General Movement Assessment

Among the six videos returned from one family, the first one from each of the two days was selected for GMA analysis. Exclusion of the remaining 4 videos and additional 4 videos excluded due to PTA outside of the age required for assessment of FMs gave a total of 122 videos available for quality assessment.

One hundred and twenty-one (99%) out of 122 videos which were returned within the required time window were classified as GMA scorable. The video that was non-scorable had infant in side lying position. Details about compliance to the In-Motion standards are shown in Table 2.

Table 2 Compliance to In-Motion standards (n= 126)

	Active movements (not hypokinetic)	Supine position	Correct state	No disturbances during recording	Adequate clothing	Adequate light	Whole body visible	Feet of parents visible
N (%)	126 (100)	125 (99.2)	122 (96.8)	124 (98.4)	125 (99.2)	124 (98.4)	124 (98.4)	116 (92.1)*
		Optimal stability	Abrupt displacement	Predominantly unstable	Correct base of support		Image quality	
						Clear	Blurred	Very blurred
N (%)		80 (63.5)	26 (20.6)	22 (17.5)	119 (94.4)	114 (90.5)	11 (8.7)	1 (0.8)

*3 missing data.

3.3 General Movement Assessment

Of the 121 videos classified as GMA scorable, 3 (2.4%) videos were classified with exaggerated (Fa), 3 (2.4%) with absent (FM-), and 7 (5.6%) with sporadic (FM+/-) FMs. Eighty-seven (69%) and 21 (16.7%) videos were classified with intermittent (FM+) and continuous (FM++) FMs, respectively.

3.4 Computer-based body point tracking

The proportion of correctly predicted left wrist key point from 5493 tested video images was 83.15%. Details of accuracy of 7 predicted body points and mean value for all points are shown in Figure 2.

3.5 Parent responses

Survey responses were received from 64 (92.8%) families of the 69 families who returned at least one video. Fifty-four (84.3%) of them observed the instructional video one or two times before filming their baby. No families returned a video without training on filming their baby first.

The majority of the survey respondents found the In-Motion-App easy to use. All respondents agreed or strongly agreed that it was easy to understand how to stand and hold the smartphone during the filming. Details about family responses are shown in Table 3. Fifty-seven (90.5%) families strongly disagreed, disagreed or neither disagreed nor agreed that they did become more worried about their child's development through using the In-Motion instructions.

Table 3 Parents' responses to the In-Motion-App and instructional video

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree
In-Motion_App					
The In-Motion app was generally easy to use	58.7%	33.3%	3.2%	4.8%	0.0%
It was easy to enter the information needed in the In-Motion app	46.0%	41.3%	11.1%	1.6%	0.0%
The reminders about when the child should be filmed were helpful	44.4%	41.3%	6.3%	6.3%	1.6%
The number of reminders about when the child should be filmed was suitable	25.4%	39.7%	15.9%	14.3%	4.8%
There were no technical problems with uploading and sending the videos	55.6%	22.2%	6.3%	4.8%	11.1%
In-Motion standards for remote GMA					
It was easy to understand how I should stand and hold the telephone during the filming	60.0%	40.0%	0.0%	0.0%	0.0%
It was easy to keep the telephone still while I was filming	16.7%	46.7%	30.0%	6.7%	0.0%
It was easy to do the filming without disturbing the child	36.5%	42.9%	9.5%	11.1%	0.0%
It was easy to understand how my child should be dressed when filmed	65.1%	33.3%	0.0%	1.6%	0.0%
It was easy to understand how my child should be positioned and how the mat should be when I was going to film	63.5%	28.6%	4.8%	3.2%	0.0%
It was easy to follow the instructions about how the lighting should be during the filming	36.5%	38.1%	14.3%	11.1%	0.0%
Filming my child for 3 minutes went smoothly	39.7%	39.7%	19.0%	1.6%	0.0%
Parental worries					
I felt safe about uploading video of my child	63.5%	27.0%	7.9%	1.6%	0.0%
I became more worried about my child's development through using the In-Motion app	1.6%	7.9%	28.6%	22.2%	39.7%
Using the In-Motion app made me more attentive to my child's development	7.9%	36.5%	46.0%	4.8%	4.8%

DISCUSSION

In this study, more than 95% of families with high-risk infants filming their baby at home, returned at least one video that was in accordance with the In-Motion standards for remote GMA. Most families found the In-Motion-App easy to use and the instructions for filming easy to follow, and less than 10% of respondents became worried through using the In-Motion-App. Despite the use of hand-held smartphones introducing movement artifacts in the video image, our computer-based 7-point body tracker detected positions of the body points with high accuracy. To the best of our knowledge, this is the first automatic infant body point tracker that is tested on video recordings from hand-held smartphones.

This study has several strengths. First, it included families of high-risk infants frequently seen in NICU follow-up settings. We argue that this makes our findings robust and generalizable to comparable clinical settings. Second, the communication of In-Motion standards through a simple and short animated video, makes the instructions easily applicable to a broad range of different clinical settings. Furthermore, the accuracy of the 7-point body tracker software with the use of smartphone videos makes it pioneering in the field of automatic and markerless infant motion capture compared to other studies (19, 20). This facilitates further development of methods for early automated detection of CP based on smartphone videos. Finally, the design of the study using an experienced and certified GMA observer for evaluation of video quality and a survey with very high response rate, makes the study quality high and the results trustworthy.

There are also several limitations. First, almost 20% of the included families did not return any video. This study was not designed to evaluate reasons for not recording or returning videos. We can, therefore, only conclude on the quality of returned videos. The questions in our survey may also have limitations, mainly covering topics favorable to participants returning videos, participating in follow-up and smartphone usage, giving little or reduced information about responders with low mobile health technology usage. Hence, problems encountered by families who did not record or return any video need to be further explored. Our findings are in accordance with the study by Kwong et al (11), where 24% of families did not return any video using the Baby Moves App. These findings indicate that home-based video recordings for remote GMA is not a solution for all families and that it might be difficult in a clinical setting to know beforehand which families will return a video or not. Furthermore, 13 (19%) families returned one or several videos outside the required time window needed for a valid GMA. More than 90% of respondents found the reminders in the App helpful, but one fifth disagreed that the number of reminders were appropriate. These findings indicate a limitation in the design of the app reminders and lack of programmed filming windows parameters. These app functionalities need to be improved in a process involving the users.

Second, our study comprised five different hospital sites in three different countries, and information provided to families when downloading the App may have differed. Additionally, there is a risk that the research personnel could have given more information and assistance to families than will be common in an ordinary clinical setting. Our study setting could therefore be in slight contrast to an ordinary clinical setting, where adapted and flexible family information is needed due to challenges in reduced participation in neurodevelopmental follow-up (21) and racial and socioeconomic differences in mobile health technology usage (22).

Third, even though almost 20% of the hand-held videos in our study were classified as predominantly unstable, the GMA expert considered the videos to be GMA scorable. There is a risk that more videos might have been classified as non-scorable by another GMA observer with less training and experience or if there had been several GMA experts observing the same videos. To the best of our knowledge, there is only one study protocol planning to assess the predictive validity of

GMA from videos taken with hand-held smartphones for CP outcome (10). Hence, further studies on the use of smartphones for GMA are needed.

Finally, our computer-based 7-point body tracker showed accurate estimations compared to manual annotations on the video image. However, further studies must explore how selection of body points, tracked body point accuracy and movement artifacts in camera will influence a machine-learning model for prediction of CP from smartphone video recordings.

This study facilitates and contributes to the use of smartphone technology for video recordings and remote GMA. Consequently, it will contribute to giving high-risk infants and their families equal access to GMA as an accurate method for early identification of CP, without geographical constraints. The use of early remote medical assessment will improve the organization and resources used in follow-up screening at hospitals and have the potential to reduce medical complications for children with CP due to early detection. A clinical feasible computer-based movement analysis with equal accuracy as GMA, will greatly reduce the need for specialized GMA observers and provide an innovative resource-effective diagnostic measure.

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Conceptualization; LA, RS, HP, OMS, TF, SO, WS, BE, CB, Data curation; LA, RS, EI, DG, Formal analysis; LA, RS, EI, DG, Funding acquisition; LA, RS, Methodology; LA, RS, EI, DG, TF, SO, Project administration; LA, BE, HP, AP, AB, CB, RS, KD, Resources; LA, TF, SO, AB, AP, HP, OMS, BE, WS, KD, Writing-original draft; LA, RS, EI, DG, Writing-review & editing; LA, RS, EI, DG, TF, SO, BE, WS, AB, AP, CB, HP, OMS, KD.

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Competing interests

None declared.

Patient consent

Obtained in all participants.

Ethics approval

All infant/families provided written informed consent and ethics was approved by the regional committee for medical and health research ethics (REC Central-Committee 2017/913) in Norway. The study sites in Denmark and Belgium also had approvals from their local Institutional Review boards. Study registration in ClinicalTrials.gov Protocol Record 2017/913.

Provenance and peer review

Not commissioned; external peer reviewed.

Data availability statement

No data are available. Data from this study are not available for sharing due to ethical approval requirements. Researchers interested in collaboration should contact the corresponding author with their expression of interest.

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Figure title – Figure 1

Figure 1: Screenshot of In-Motion instructional video showing examples of information about infant state, lighting and positioning of baby and person filming the baby.

Figure title – Figure 2

Figure 2 Accuracy of 7 estimated body points compared with manually annotated images

Figure 2: From left: Table with proportions of correct detected body point; illustration of the computer-based detections according to 7 body points; the distribution of the left wrist body point detections (blue dots) relative to the manually annotated landmark (black dot) where 10% of the infant head size is used as a threshold (black circle).

Figure title and legend – Figure 3

Figure 3 Flow chart of infant/families with reasons for non-upload of videos.

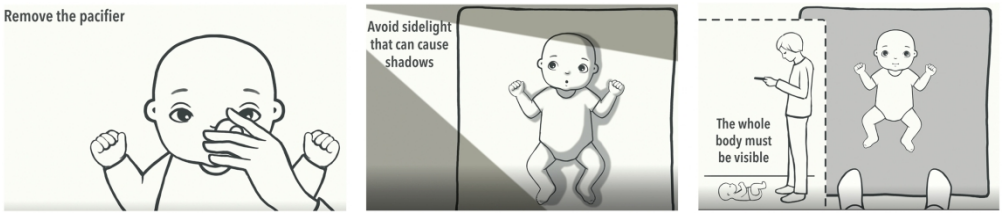
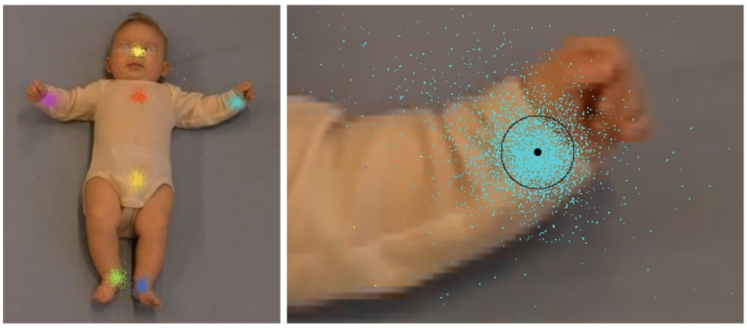


Figure 1: Screenshot of In-Motion instructional video showing examples of information about infant state, lighting and positioning of baby and person filming the baby.

209x44mm (300 x 300 DPI)

	%
Mean	80.9
Nose	93.3
Thorax	93.9
Right wrist	74.4
Left wrist	83.1
Pelvis	63
Right ankle	79.5
Left ankle	79.3



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209x68mm (300 x 300 DPI)

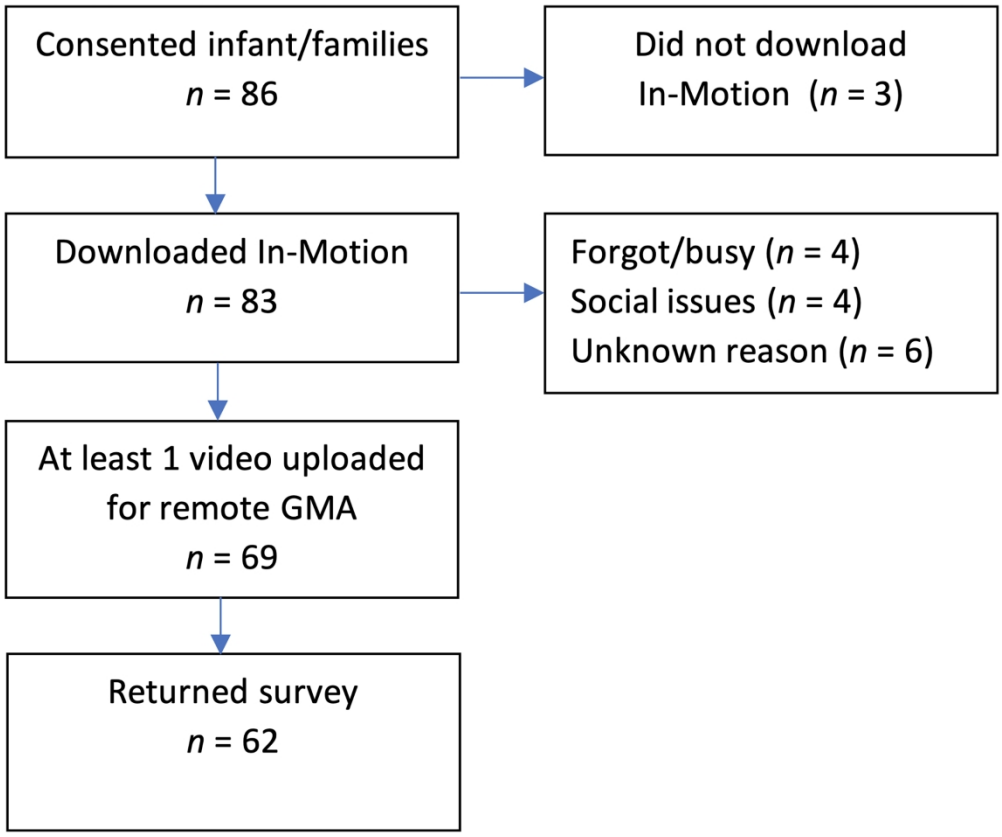

Figure 3 Flow chart of infant/families with reasons for non-upload of videos.

209x174mm (300 x 300 DPI)

Appendix S1 – Parent survey

Thank you for taking the time to answer these questions. It takes about 10-15 minutes to answer them. If you have questions, you can contact …, email ……, or mobile telephone …….

1. Are you the child's mother or father?

- Mother
- Father

2. What is your marital status?

- Single
- Married/cohabitant
- Separated/divorced
- Widow/widower

3. What is your age and what is the age of your spouse/partner?

- You:
- Your spouse/partner:

4. How many brothers and sisters live with the child sometimes or always?

5. What type of mobile phone do you have?

- iPhone (iOS/Apple)
- Android

6. To what extent do you agree or disagree with each of these statements?

The In-Motion-App was generally easy to use

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to enter the information needed in the In-Motion-App

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

The reminders about when the child should be filmed were helpful

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

The number of reminders about when the child should be filmed was suitable

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

There were no technical problems with uploading and sending the videos

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to understand how I should stand and hold the telephone during the filming

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to keep the telephone still while I was filming

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to do the filming without disturbing the child

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to understand how my child should be dressed when filmed

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to understand how my child should be positioned and how the mat should be when I was going to film

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

It was easy to follow the instructions about how the lighting should be during the filming

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

Filming my child for 3 minutes went smoothly

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

I felt safe about uploading video of my child

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

I become more worried about my child's development through using the In-Motion-App

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

Using the In-Motion app made me more attentive to my child's development

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

STROBE 2007 (v4) Statement—Checklist of items that should be included in reports of *cohort studies*

Section/Topic	Item #	Recommendation	Reported on page #
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract	1
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found	2
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	3
Objectives	3	State specific objectives, including any prespecified hypotheses	3
Methods			
Study design	4	Present key elements of study design early in the paper	3
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	4
Participants	6	(*a*) Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up	4
		(*b*) For matched studies, give matching criteria and number of exposed and unexposed	na
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	4-5
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	4-5
Bias	9	Describe any efforts to address potential sources of bias	5
Study size	10	Explain how the study size was arrived at	4
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	6
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding	6
		(*b*) Describe any methods used to examine subgroups and interactions	na
		(*c*) Explain how missing data were addressed	na
		(*d*) If applicable, explain how loss to follow-up was addressed	na
		(*e*) Describe any sensitivity analyses	na
Results			

Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	6
		(b) Give reasons for non-participation at each stage	6
		(c) Consider use of a flow diagram	6
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	7
		(b) Indicate number of participants with missing data for each variable of interest	na
		(c) Summarise follow-up time (eg, average and total amount)	na
Outcome data	15*	Report numbers of outcome events or summary measures over time	6-7
Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	na
		(*b*) Report category boundaries when continuous variables were categorized	na
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	na
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	6-7
Discussion			
Key results	18	Summarise key results with reference to study objectives	7
Limitations			
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	7
Generalisability	21	Discuss the generalisability (external validity) of the study results	7-8
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	8

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at www.strobe-statement.org.