



Are sporadic fidgety movements as clinically relevant as is their absence?



Christa Einspieler^{a,*}, Hong Yang^b, Katrin D. Bartl-Pokorny^a, Xia Chi^c, Fei-Fei Zang^b, Peter B. Marschik^{a,d}, Andrea Guzzetta^{e,f}, Fabrizio Ferrari^g, Arend F. Bos^h, Giovanni Cioni^{e,f}

^a Research Unit iDN, Interdisciplinary Developmental Neuroscience, Institute of Physiology, Center for Physiological Medicine, Medical University of Graz, Austria

^b Department of Rehabilitation, Children's Hospital of Fudan University, Shanghai, PR China

^c Department of Pediatrics, Nanjing Maternity and Child Health Hospital, Nanjing Medical University, PR China

^d Center of Neurodevelopmental Disorders, Karolinska Institutet, Stockholm, Sweden

^e Department of Developmental Neuroscience, University of Pisa, Italy

^f IRCCS Fondazione Stella Maris, Pisa, Italy

^g Dept. of Neonatology, University of Modena and Reggio Emilia, Modena, Italy

^h Division of Neonatology, Beatrix Children's Hospital, University Medical Center, University of Groningen, The Netherlands

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ABSTRACT

Background: Infants with normal fidgety movements at 3 to 5 months after term are very likely to show neurologically normal development, while the absence of fidgety movements is an early marker for an adverse neurological outcome, mainly cerebral palsy (CP). The clinical significance of so-called sporadic fidgety movements (i.e., fidgety movements occur isolated in a few body parts and are of 1- to 3-second-duration) is not yet known.

Aims: Our objective was to determine whether infants who had developed CP and had sporadic fidgety movements have a better outcome than infants who did not have fidgety movements.

Study design: Longitudinal study. Retrospective analysis of prospectively collected data.

Subjects: 61 infants who developed CP (46 male, 15 female; 29 infants born preterm; videoed for the assessment of movements and postures at 9 to 16 weeks post-term age).

Outcome measures: The Gross Motor Function Classification System (GMFCS) was applied at 3 to 5 years of age. **Results:** There was no difference between children diagnosed with CP who had sporadic fidgety movements at 9 to 16 weeks post-term age ($n = 9$) and those who never developed fidgety movements ($n = 50$) with regard to their functional mobility and activity limitation at 3 to 5 years of age. One infant had normal FMs and developed unilateral CP, GMFCS Level I; the remaining infant had abnormal FMs and developed bilateral CP, GMFCS Level II.

Conclusions: There is no evidence that the occurrence of occasional isolated fidgety bursts indicates a milder type of CP.

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1. Introduction

After the assessment of general movements (GMs) was introduced [1], its main field of application has been the prediction of cerebral palsy (CP) [2,3]. Apart from abnormal cramped-synchronised GMs around term age [1,4], it is particularly the absence of fidgety GMs at 3 to 5 months after term that is an early marker for CP [1,5]. Fidgety movements (FMs) are typically of small amplitude and moderate speed with a variable acceleration of small movements of the neck, trunk and limbs in all directions in the awake infant, except during fussing and crying [1,6]. They may be found as early as 6 weeks post-term but usually occur at around 9 weeks and are present until an age of 20 weeks or even a few weeks longer, by which time intentional

and antigravity movements occur and start to dominate [1,6,7]. This age range holds true for both term and preterm infants after correcting the age [8–10]. The temporal organisation of FMs varies with age: initially, they occur as isolated events before gradually increasing in frequency until finally, by the age of 16 to 20 weeks, they subside [11]. Gross movements such as kicking, swipes, wiggling–oscillating arm movements, movements to the midline or antigravity movements may occur together with FMs. That is to say that FMs are superimposed on other movements or other movements may occur during the pauses between FMs, or both [7].

In typically developing 9- to 15-week-old infants continual FMs (score: F++) or intermittent FMs (score: F+) are obligatory [10–13]; for definitions and illustration see Table 1 and Fig. 1. Usually, the temporal organisation of FMs is rather robust and could not be significantly changed by different kinds of manipulation such as presenting visual or acoustic stimuli, approach of the caregiver, or hemi-loading of the infant [12,13]. Only the presentation of high-contrast faces caused a decrease or stop of FMs though for no longer than 20s [12].

* Corresponding author at: Research Unit iDN, Interdisciplinary Developmental Neuroscience, Institute of Physiology, Medical University of Graz, Harrachgasse 21, 8010 Graz, Austria. Tel.: +43 316 380 4266; fax: +43 316 380 9630.

E-mail address: christa.einspieler@medunigraz.at (C. Einspieler).

Table 1
Temporal organisation of fidgety movements (FMs) [11–13].

Classification	Score	Definition
Continual FMs	F++	FMs occur frequently but are interspersed with very short (i.e. 1–2 s) pauses (Fig. 1, line C). As FMs are by definition GMs, they involve the whole body, particularly the neck, shoulders, wrists, hip, and ankles. Depending on the actual body posture, especially on the position of the head, FMs may occur asymmetrically. If the infant is focused on the environment, his or her FMs are mainly displayed in the hips and ankles, and are less obvious in the shoulders and wrists.
Intermittent FMs	F+	Although FMs occur in all body parts, the temporal organisation differs from F++. Here, the pauses between FMs are prolonged (1–10 s), which creates the impression that FMs are only present during half of the observation time (Fig. 1, line B).
Sporadic FMs	F+/-	Sporadic FMs (Fig. 1, line A) are interspersed with long pauses (up to 1 min). FMs may occur isolated in a few body parts and are of very short duration (1 to 3 s).
Absence of FMs	F-	No FMs can be observed, although other movements may occur.

Infants with normal intermittent or continual FMs are very likely to show neurologically normal development, even if they belong to a high-risk group for maldevelopment. Several large-scale studies reported sensitivities of 95% to 100% and specificities of 96% to 98% for the assessment of FMs [1,14,15]. If FMs are totally absent at 3 to 5 months (score: F-), the infant has a high risk for neurological impairments, mainly spastic uni- or bilateral CP [1,5,14–19], but also dyskinetic CP [20]. Apart from the absence of FMs, a cramped-synchronised movement character, repetitive opening and closing of the mouth as well as abnormal finger postures are more common in infants who later develop CP with severe functional limitations [18,19] as classified on the Gross Motor Function Classification System (GMFCS) [21]. By contrast, a normal posture, absent FMs, and an abnormally jerky but neither monotonous nor cramped-synchronised overall movement character were identified in infants who were later classified at GMFCS Levels I or II (i.e. mild functional limitations) [18,19].

During the second month after term FMs may occur sporadically, i.e. as brief and isolated events. At 3 to 4 months, however, such sporadic FMs (score: F+/-; Table 1, Fig. 1A) are considered age-inadequate. In fact most of the researchers included 3- to 4-month-old infants with sporadic FMs in the group of absent FMs, without either mentioning such a distinction or giving it any further attention [22,23]. Mutlu et al. [22] reported that the almost full agreement between three scorers on 30 individuals assessed three times was slightly reduced as one infant (recorded at 12 weeks) was inconsistently scored by one observer, and this disagreement was due to sporadic vs. absent FMs. The authors discussed that such a disagreement in inter-rater and intra-individual reliability would be irrelevant in a clinical setting “as sporadic FMs would have been as much a cause of concern as the absence of FMs” [22, p. 216]. Hamer et al. [24] described sporadic FMs in ten out of 44 high-risk-infants, of whom only one individual was diagnosed with (unilateral) CP, GMFCS Level II. The remaining nine individuals with sporadic FMs were annotated as no-CP at the 18-month-outcome assessment. However, the authors alluded to the fact that 18 months is relatively early for determining the functional and cognitive outcomes [24]. Therefore, it still remains to be seen if a distinction between absent and sporadic FMs is of clinical significance.

In order to shed light on this blind spot, we re-assessed our prospectively collected footage of infants later diagnosed with CP, and paid special attention to the temporal organisation of their FMs. The aims of our study were (1) to elaborate on sporadic FMs (i.e. duration of single bursts, interval duration between bursts); (2) to analyse the extent to which the temporal organisation of FMs was associated with the concurrent motor repertoire; and (3) to analyse to what extent the temporal organisation is related to the functional mobility and activity limitation at 3 to 5 years of age as classified on the GMFCS.

2. Methods

2.1. Participants

The study comprised 61 children – 46 boys (75.4%) and 15 girls (24.6%) – who had been admitted to (a) the Department of Rehabilitation at the Children's Hospital of the Fudan University, Shanghai, or (b) the Department of Pediatrics, Nanjing Maternity and Child Health Hospital, Nanjing Medical University, PR China between September 2003 and June 2010, and had been diagnosed with spastic CP at the age of 2 to 3 years. The reasons for admission were the following: (a) a high risk for neurodevelopmental disorders due to preterm birth or perinatal asphyxia at term; (b) abnormal findings at paediatric examinations; or (c) parental concerns. Some of the participants had also been included in a previous study [19]. The inclusion criteria for this study were (i) that their motor performance had been videoed at around 4 months of age and (ii) that their gross motor function had been classified by means of the GMFCS [21] at 3 to 5 years of age. Table 2 provides the clinical characteristics of the participants.

2.2. Procedure and assessments

Five- to 7-minute video recordings were made prospectively of the spontaneous motility of each infant at 9 to 16 weeks post-term age. The recordings were made during periods of active wakefulness between feedings, with the infant dressed in a bodysuit, lying in supine position [25]. The video recordings were evaluated retrospectively by

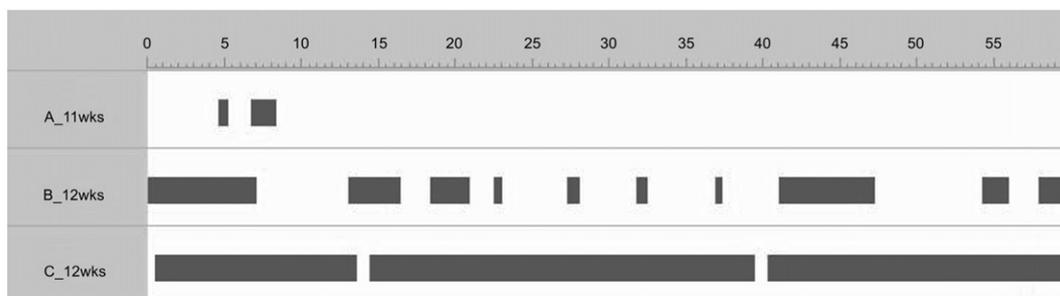


Fig. 1. Temporal organisation of FMs; the duration of the actograms is 60 s. Line A = 11-week-old infant with sporadic FMs (score F+/-); line B = 12-week-old infant with intermittent FMs (score: F+); line C = 12-week-old infant with continual FMs (score: F++).

Table 2

Clinical characteristics of 61 children who were diagnosed with cerebral palsy at 2 to 3 years of age, according to the quality and temporal organisation of their FMs. Statistical differences refer to sporadic vs. absent FMs. Data on the infant with normal intermittent FMs and the infant with abnormal FMs are merely descriptive.

	Normal FMs (n = 1)	Abnormal FMs (n = 1)	Sporadic FMs (n = 9)	Absent FMs (n = 50)	p
Male:female	1:0	0:1	8:1 (89%:11%)	37:13 (74%:26%)	0.334 ^a
Preterm birth	0	1	7 (78%)	21 (42%)	0.048^a
Gestational age in weeks	38	35	Md = 31 IQR = 28–36	Md = 37 IQR = 32–39	0.039^b
Birthweight in grams	4200 g	2050 g	Md = 1340 IQR = 1170–2497	Md = 2956 IQR = 1889–3313	0.028^b
Periventricular leucomalacia	–	1	3 (33%)	10 (20%)	0.285 ^a
Intracranial haemorrhage	1	–	2 (22%)	18 (36%)	0.519 ^a
Hypoxic ischaemic encephalopathy	–	–	1 (11%)	8 (16%)	0.783 ^a
Normal brain image ^c	–	–	1 (11%)	4 (8%)	
Outcome at 3–5 years of age					
Unilateral spastic CP	1	–	–	3 (6%)	
Bilateral spastic CP	–	1	9 (100%)	47 (94%)	
GMFCS ^{Ref}					0.072 ^b
Level 1	1	–	5 (56%)	9 (18%)	
Level 2	–	1	–	6 (12%)	
Level 3	–	–	1 (11%)	7 (14%)	
Level 4	–	–	2 (22%)	14 (28%)	
Level 5	–	–	1 (11%)	14 (28%)	

Key: Md = median.

^a Pearson chi-square test.

^b Mann–Whitney U test.

^c Brain imaging was performed in 56 infants (92%) by means of magnetic resonance (n = 11), cranial ultrasound or computer tomography.

at least three scorers (HY, XC, CE, KBP, PBM, all of whom are certified GM assessors) according to the Precht method of GM assessment [11]. Three scorers (CE, KDB, PBM) were not familiar with the participants' clinical outcomes and histories. In case of disagreement (in 5 recordings; 8%), the scorers re-evaluated the recording of the infant in question until a consensus was reached on a final score.

FMs and the concurrent motor repertoire (23 movement patterns and 13 postural patterns) were assessed independently in separate runs from the video recordings. The temporal organisation of FMs was

coded by KDB and CE using the software Noldus Observer-XT (www.noldus.com). This software allows coding observational data in millisecond intervals and offers the possibility of visualising the duration of each burst of FMs by tagging onset and offset for each FM event (Fig. 2). The coding resulted in the calculations of (i) the duration of FM bursts and (ii) the duration of intervals between bursts.

Using the score sheet for the assessment of the motor repertoire at 3 to 5 months [11, p. 26], we calculated a Motor Optimality Score with a maximum value of 28 (the best possible performance) and a minimum

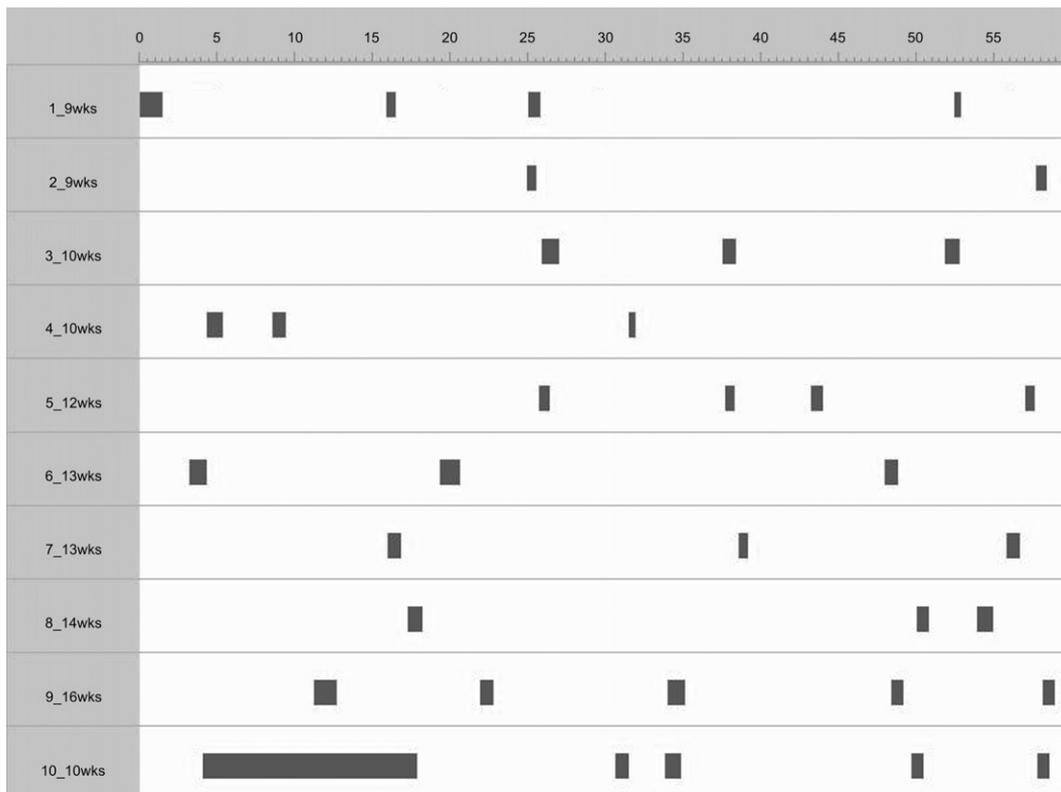


Fig. 2. Temporal organisation of FMs in study cases who showed either sporadic or intermittent FMs. The duration of the actograms is 60 s. Lines 1 to 9 depict actograms of the nine infants with sporadic FMs (score F+/-); line 10 illustrates the actogram of the infant with intermittent FMs (score: F+). Key: wks = weeks post-term age.

value of 5. The score sheet comprises five sub-categories: (i) FMs; (ii) age-adequacy of motor repertoire; (iii) quality of movement patterns other than FMs such as wiggling-oscillating arm movements, swipes, kicking, head and mouth movements, movements towards the midline (hand-to-hand, foot-to-foot, hand-to-mouth), legs lift, rolling, etc.; (iv) posture such as head in midline, symmetrical body posture, asymmetric tonic neck response, finger postures; and (v) overall movement quality (for details see [19,26,27]). A reliability study revealed intra-class correlation coefficients ranging between 0.80 and 0.94 [28].

Diagnosis of CP was carried out at >30 months post-term age and was based on recent definitions and consensus criteria [29,30]. Four children (6.5%) were diagnosed with unilateral spastic CP. Bilateral spastic CP was diagnosed in 57 children (93.5%), whereby the arms were less affected than the legs in 19 children. Epilepsy was diagnosed in 11 children (18%); severe visual impairment in 12 children (20%); and severe hearing loss was found in 6 children (10%). Data on cognitive development are not available.

At 3 to 5 years, all children were scored and classified by two rehabilitation doctors (HY, XC) according to the GMFCS [21]. The GMFCS is a standardised method of classifying the gross motor function of children with CP. It is based on a five-level classification system: the higher the level, the more severe the CP (Table 2).

The study has been approved by the Institutional Review Boards of the Children's Hospital of the Fudan University, Shanghai, and the Nanjing Medical University, Nanjing, PR China.

2.3. Statistical analysis

Statistical analysis was performed using the SPSS package for Windows, version 22.0 (SPSS Inc., Chicago, IL). Pearson chi-square test and chi-square test for trend were used to evaluate associations between nominal data. The Mann-Whitney U test was used to compare the two groups when data were interval or ordinal scale. Throughout the analyses, $p < 0.05$ (two-tailed) was considered to be statistically significant.

3. Results

3.1. Fidgety movements

One infant had normal intermittent FMs (score: F+; Fig. 2, line 10); one infant had continual abnormal FMs, i.e. exaggerated in amplitude and speed [11]; nine infants (15%) displayed sporadic FMs (score: F+/-; Fig. 2, lines 1 to 9). The average duration of FM bursts was 0.82 s (SD = 0.24 s); the maximal duration was 2.86 s. The intervals

between FM bursts were 3 to 47 s (mean = 17.2 s, SD = 9.2 s). The other 50 infants (82%) showed no FMs at all (score: F-).

3.2. Association between FMs and the concurrent motor activity

The motor repertoire was scored as reduced in 5 infants (8%), and not adequate to age in 56 infants (92%). Infants with sporadic FMs scored significantly better than infants with absent FMs although none of the infants displayed an age-adequate movement repertoire ($p < 0.05$; Table 3). Movement patterns (concurrent to FMs) were scored as predominantly normal in 5 infants (8%) and predominantly abnormal in 42 infants (69%); the remaining 14 infants (23%) had an equal number of normal and abnormal movement patterns. Posture was rated as predominantly normal in 9 infants (15%) and predominantly abnormal in 44 infants (72%); eight infants (13%) showed an equal number of normal and abnormal postural patterns. Neither movement patterns nor postural patterns were related to the temporal organisation of FMs (n.s., Table 3).

None of the infants had a normal smooth and fluent movement character. Cramped-synchronised movements were observed in 18 infants (29.5%), none of whom had FMs ($p < 0.05$; Table 3). Forty-three infants (70.5%) showed a monotonous, jerky or tremulous movement character.

The median motor optimality score was 6 (P25 = 6, P75 = 8, range = 5–20), but infants with sporadic FMs did not differ from infants whose FMs were absent (n.s.; Table 3). The motor optimality scores of infants with normal vs. abnormal FMs were 20 and 16, respectively (Table 3).

3.3. Association between FMs and the functional mobility and activity limitation at age 3 to 5 years

Outcome assessments revealed GMFCS Level I in 15 children (25%; including three with unilateral spastic CP), Level II in seven children (11.5%; one child with unilateral spastic CP), Level III in eight children (13%), Level IV in 16 children (26%), and Level V in 15 children (24.5%). The distribution of GMFCS Levels was the same across the categories of absent vs. sporadic FMs (n.s.; Table 2). Comparing the temporal organisation of FMs in individuals later diagnosed with GMFCS Levels I or II vs. Levels III, IV or V did not reveal any difference ($p = 0.130$). Whether FMs are absent or sporadically present was not associated with the later ability to ambulate. The child with normal FMs developed unilateral CP, GMFCS Level I. The child with abnormal FMs developed bilateral CP, GMFCS Level II.

Table 3
Movements and postures in 61 infants at 9 to 18 weeks post-term age, according to the quality and temporal organisation of their FMs. Statistical differences refer to sporadic vs. absent fidgety movements. Data on the infant with normal intermittent fidgety movements and the infant with abnormal fidgety movements are merely descriptive.

	Normal FMs (n = 1)	Abnormal FMs (n = 1)	Sporadic FMs (n = 9)	Absent FMs (n = 50)	p
Movement repertoire adequate:reduced:not adequate	Reduced	Reduced	0:2:7	0:1:49	0.012^a
Quality of movement patterns N > A:N = A:N < A ^c	N = A	N > A	2:2:5	2:11:37	0.062 ^a
Quality of postural patterns N > A:N = A:N < A ^c	N = A	N > A	0:1:8	8:6:36	0.194 ^a
Movement character N:A but not CS:CS	Monotonous	Monotonous	0:9:0	0:32:18	0.032^a
Motor optimality score: median (IQR), range	20	16	7 (6–8) range: 6–10	6 (5–7) range: 5–10	0.119 ^b

Key: A = abnormal; CS = cramped-synchronised, i.e. movements appear rigid, all limb and trunk muscles contract almost simultaneously and relax almost simultaneously [11]; IQR = interquartile range; N = normal.

^a Chi-square test for trend; $p < 0.05$ emphasised in bold.

^b Mann-Whitney U test.

^c N > A (predominantly normal); N = A (equal number of normal and abnormal patterns); N < A (predominantly abnormal)

4. Discussion

Due to the application of event logging software, this was the first study to provide a numeric description of the temporal organisation of FMs, particularly of sporadic FMs. None sporadic fidgety burst lasted longer than 3 s. The average duration of less than 1 s may therefore explain the difficulty of reliably distinguishing between sporadic and absent FMs [22], especially if we consider that the intervals between those bursts can last up to 1 min.

Our analysis further demonstrated that infants with sporadic FMs and those with absent FMs (who developed CP) showed no difference with respect to their GMFCS Levels 3 to 5 years later. However, sporadic FMs were linked to a slightly better (although not normal) concurrent movement repertoire. Moreover, none of the infants with sporadic FMs had a cramped-synchronised movement character (Table 3), which confirms that infants who still have cramped-synchronised movements at 3 to 4 months of age are unable to perform any fidgety activity [11,31], be it only sporadically. But these differences did not express themselves in terms of significantly different motor optimality scores between both groups. In this context, it is worthwhile mentioning that an ongoing American-Norwegian study [32] has found a slightly higher motor optimality score where FMs were sporadically present as opposed to infants without FMs, although their sample was relatively small (11 infants with sporadic FMs vs. 15 infants without FMs). We should also note that this study was performed in a medical complex population with brain injuries, surgical congenital heart diseases and prolonged hospitalisation [32].

One of our study cases had normal intermittent FMs and developed unilateral CP, GMFCS Level I. Her neonatal cranial ultrasound revealed cystic periventricular leucomalacia (Table 2). The observation of normal FMs is in line with previous reports of a few children who had normal FMs and developed CP, albeit in its mild form [1,24,26,27].

Another infant had abnormally exaggerated FMs and was later diagnosed with bilateral CP, GMFCS Level II. His neuroimaging revealed intracranial haemorrhage with parenchymal involvement (Table 2). Still, the significance of abnormal FMs is not clear. This motor abnormality was more often reported in infants who developed minor neurological dysfunctions at school age [33] and was recently also discussed as an early marker for autism spectrum disorder [34]. By and large, the predictive value of abnormal FMs remains low as the outcomes ranges from normal to CP [1,6].

4.1. Limitations

Admittedly, our sample of nine infants with sporadic FMs was small. Also, our study group may not be representative of the general CP population; only four individuals were diagnosed with unilateral CP. Moreover, we only focused on GMs in infants who were later diagnosed with CP, and we only used the GMFCS as an outcome measure. In order to further elucidate the clinical relevance of sporadic FMs, we would need to know their frequency of occurrence in the normal population. So far, only one major study has been conducted on the prevalence of abnormal GMs in 455 3-month-old infants representing the general population [35]. But the study in question did not focus on FMs and, least of all, their temporal organisation but classified GMs according to Hadders-Algra as normal optimal, normal suboptimal, mildly abnormal and definitely abnormal [2]. For the moment we can therefore only draw conclusions from a small sample of infants with sporadic FMs developing CP.

5. Conclusion

Our study indicates that 15% of the infants who later developed CP had sporadic FMs which was linked to a slightly better (although not normal) concurrent movement repertoire. At any rate, the presence of a few isolated fidgety bursts lasting up to 3 s in high-risk 3- to

4-month-old infants who were later diagnosed with CP did not prove diagnostically conclusive for later functional mobility and activity limitations. In other words, we still do not know if sporadic FMs are of any clinical relevance in infants who do not develop CP but are at a certain risk for an adverse neurodevelopmental outcome.

Conflict of interest

None to declare.

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