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Are abnormal fidgety movements an early marker for complex minor neurological dysfunction at puberty?

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Follow-up;
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Abstract

Background: Prechtl's method on the qualitative assessment of general movements (GMs) is a powerful tool for early and specific prediction of cerebral palsy. However, it is uncertain whether the GM assessment can be used to predict mild neurological impairment.

Aims: To determine whether the quality of general movements (GMs) from the age of 3 to 5 months, i.e. fidgety movements, is related to the presence of complex minor neurological dysfunctions (MND) 13 to 15 years later.

Study design: Prospectively collected data on the quality of GMs during infancy were retrospectively analysed on the basis of MND at puberty.

Subjects: Twenty-eight participants (14 girls and 14 boys) with a median gestational age of 40 weeks (range: 35 to 42 weeks) and an appropriate birth weight (median 3390 g; range 1900 to 4200 g).

Outcome measures: Touwen's neurological examination.

Results and conclusions: Abnormal fidgety movements were not related to later complex MND, but to fine manipulative disabilities ($p < 0.05$). Normal fidgety movements, which are continually present in the whole body, might be required for optimal calibration of the proprioceptive system.

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1. Introduction

Prechtl's method on the qualitative assessment of general movements (GMs) is a powerful tool for early and specific prediction of cerebral palsy [1,2]. However, it is uncertain

whether the GM assessment can be used to predict mild neurological impairment [3]. GMs emerge in the fetus at the postmenstrual age of 9 to 10 weeks and persist until the age of 6 months [2]. Normal GMs involve the whole body in a variable sequence of arm, leg, neck and trunk movements. They wax and wane in terms of intensity, force and speed; they also commence and end gradually. Rotations along the axis of the limbs and slight changes in the direction of movements make them fluent and elegant, and create an impression of complexity and variability [1,3]. At the end of the second month of life GMs change their character from a writhing to a fidgety appearance [4]. Fidgety movements are small movements of the neck, trunk and limbs, of moderate speed and variable acceleration, and executed in all directions. They occur continually in the awake infant, except when the infant frets or cries [5]. The absence of fidgety movements is a specific predictor of cerebral palsy [2,3,5].

As a neurological sign, fidgety movements may have an abnormal quality. Abnormal fidgety movements look like normal ones, but their amplitude, speed and jerkiness are exaggerated [3,5]. They are less predictive of the neurological outcome than is the absence of fidgety movements [5], but have been discussed in the context of the development of mild neurological impairments at two to three years of age [3,6–8]. Similar results were obtained by Hadders-Algra et al. [9–11], although their methodological approach differs from Prechtl's GM assessment [3]. According to Hadders-Algra [12,13], the so-called mildly abnormal GMs – irrespective of whether they are of a writhing or fidgety character – lack fluency but are still complex and variable. Mildly abnormal GMs indicate an increased risk for the development of minor neurological dysfunctions (MND) in 18-month-olds [10], and also in 4- to 12-year-old children [9–11].

To our knowledge, it is not known whether the quality of fidgety movements is also related to the neurological performance of young adolescents at puberty, an age at which the prevalence of MND seems to decline according to the data of the Groningen Perinatal Project [14,15]. Due to the age-specific nature of the nervous system, Hadders-Algra [16] made a distinction between complex MND before and after the onset of puberty. Young adolescents with complex MND exhibit mild coordination problems as well as mild fine manipulative disabilities [16]. The aim of the present study was to assess whether the quality of GMs from 3 to 5 months (i.e. the period of fidgety movements) is related to the presence of complex MND 13 to 15 years later.

2. Methods

2.1. Participants

The study group consisted of 28 participants (14 girls and 14 boys), born in Graz (Austria) from 1985 to 1990. All participants were born at term with an appropriate birth weight. Eighteen children were considered to be at high risk for sudden infant death due to infantile apnoea or an apparent life-threatening event. The relationship between infantile respiratory data and neurological outcome has been described elsewhere [17]. A further ten subjects were recruited from the Department of Paediatrics, Medical

University of Graz, and were considered to be at low risk for sudden infant death (Table 1). Brain ultrasound examinations were carried out in five participants and were considered to be normal. All children had participated in previous studies concerning normal and abnormal GMs [5,18,19].

From 2000 to 2005 all participants were examined neurologically between the ages of 13 and 15 years. All subjects had reached puberty according to the criteria applied by Lusing et al. [14], which are based on the signs of puberty given by Marshall and Tanner [20]. The assessments were carried out with the written informed consent of the parents and the children, and were in conformance with the standards prescribed by the ethics committee of Austria. The study was approved by the Austrian Ethics Commission.

2.2. Assessment of fidgety movements

At a median age of 15 weeks (range 9 to 20 weeks), all participants were videoed during active wakefulness while lying supine. The assessment was carried out by the work group for developmental physiology and developmental neurology at the Institute of Physiology, Medical University of Graz. The recordings had a duration of 6 to 14 min (median 10 min) and were performed according to the procedure described by Einspieler et al. [21]. Fidgety movements were scored normal or abnormal according to Prechtl et al. [3,5] (as described above, in the Introduction). In addition, the temporal organization of normal fidgety movements was scored as continual (occurring frequently but interspersed with short pauses; score++) or intermittent (the pauses between fidgety movements were prolonged, giving the impression that fidgety movements were present for only a half of the observation period; score+) [3]. A score of P=D was assigned when fidgety movements were equally present in the proximal and the distal portions of the body. A score of D>P indicated that fidgety activity occurred more frequently in the wrists and the ankles than in the trunk and the proximal joints. Children with more prominent fidgety movements in the neck, trunk, shoulders and hips were scored P>D [3]. The videos were coded and re-assessed by CE with a 13- to 15-year reliability of $r=0.81$. In addition, PBM, YN, and AFB assessed the videos without being aware of the outcome of the neurological examination at puberty. The inter-scorer agreement was excellent (Cohen's Kappa=0.84).

2.3. Neurological outcome

Between the ages of 13 and 15 years (median 13 years and 8 months) the subjects underwent a standardized age-specific neurological examination according to Touwen [22]. The purpose of this assessment was to detect minor deviations in neural functions. It consists of the observation of the child's motor behaviour and tests for specific neural functions. Touwen had based his interpretation on ten subsystems [22]. Currently six clusters of dysfunctional signs are used: dysfunctional muscle tone regulation, reflex abnormalities, choreiform dyskinesia, coordination problems, dysfunction in fine manipulative ability, and rare

Table 1 Clinical characteristics of the study groups

	High-risk group (N=18)	Low-risk group (N=10)	
Girls/boys	9/9	5/5	
Gestational age at birth (mean±SD in weeks)	39.5±0.9	39.7±0.7	n.s.
Birth weight (mean±SD in grams)	3362±200	3426±238	n.s.
Infantile apnoea/ALTE	15/3	–	
FMs (normal/abnormal)	13/5	5/5	n.s.
Neurological outcome (normal/complex MND)	10/8	8/2	n.s.

ALTE, apparent life-threatening event; FMs, fidgety movements; MND, minor neurological dysfunction.

miscellaneous disorders [16,23]. Based on a review of the Groningen Perinatal Project, Hadders-Algra made a distinction between simple MND and complex MND [16]. Before the onset of puberty the distinction between these forms is based on the number of clusters of dysfunction; after the onset of puberty it is based on the type of dysfunction. Young adolescents with complex MND exhibit both, mild coordination problems and a mild fine manipulative disability [16].

The neurological examination was videoed and scored by CE and SM. SM was unaware of the child's history. Reflexes, resistance against passive movements, and muscle power could not be double-checked. For all other items the inter-scorer agreement was excellent with a median Kappa of 0.82. In the event of disagreement, the video recordings were discussed with HFRP until agreement was achieved.

2.4. Statistics

Using SPSS 13.0, Fisher's Exact Test and Pearson's Chi-Square were applied to assess relationships between the nominal data of the quality of fidgety movements and the neurological outcome. The Student's *t*-Test was used to compare birth weight and gestational age between the two subgroups.

Table 2 Quality of fidgety movements at 3 to 5 months and neurological outcome at 13 to 15 years

	Complex MND (N=10)	Normal (N=18)	
Abnormal fidgety movements (N=10)	4	6	n.s.
Normal fidgety movements (N=18)	6	12	
Score++	2	10	<i>p</i> <0.05
Score+	4	2	
P=D	4	12	<i>p</i> <0.05
P>D	2	–	

Table 3 Quality of fidgety movements at 3 to 5 months and fine manipulative ability at 13 to 15 years

	Dysfunctional (N=14)	Normal (N=14)	
Abnormal fidgety movements (N=10)	8	2	<i>p</i> <0.05
Normal fidgety movements (N=18)	6	12	

A probability value less than 5% was considered to be significant.

3. Results

Ten infants had abnormal fidgety movements; half of them belonged to the low-risk group (Table 1). The remaining 18 infants had normal fidgety movements. Fidgety movements occurred either continually (N=12) or intermittently (N=6). In the majority of infants (N=16), normal fidgety movements were equally present in the distal and proximal portions of the body (P=D); two infants had more prominent fidgety movements in the neck, trunk, shoulders and hips (P>D; Table 2).

Eighteen participants were neurologically normal. Four girls and six boys (N=10) were classified as complex MND; two of these boys were considered as low risk during their first months of life (Table 1). Abnormal fidgety movements were not related to the occurrence of complex MND (Table 1). However, if normal fidgety movements were continually present (+) in all body parts (P=D) the neurological outcome was more likely normal (*p*<0.05, Table 2).

In addition to ten adolescents exhibiting a dysfunction in both, coordination and fine manipulation (i.e. the definition of complex MND), four participants had solely a fine manipulative disability without coordination difficulties. As these four participants had abnormal fidgety movements during their infancy, abnormal fidgety movements were related to a dysfunction of the cluster 'fine manipulative ability' (*p*<0.05, Table 3).

4. Discussion

In a previous study on the short-term effects of infantile respiratory problems we recorded the general movements of these infants. A strong correlation existed between the respiratory measurements and the impairment of general movements [18,19]. The long-term sequelae of infantile apnoea and idiopathic apparent life-threatening events revealed a high incidence of MND [17]. By contrast, we failed to demonstrate a relationship between general movements, in particular fidgety movements, and complex MND 13 to 15 years later. Ten infants had abnormal fidgety movements; but, only four of them were subsequently assigned to the category of complex MND. The remaining six subjects were neurologically normal after the onset of puberty (Table 2). This confirmed the low predictive value of abnormal fidgety movements [2,3,5]. Only Bos et al. [6–8] observed a higher incidence of abnormal fidgety movements in children with MND at the preschool age. However, data collected at the preschool age cannot be extrapolated to puberty, as the

prevalence of MND is known to be age-specific and decreases with the onset of puberty [14,15,23]. In respect to normal fidgety movements, their continual presence all over the body (score:++, P=D) was more likely related to later normal neurological development than intermittent fidgety movements. This confirms the clinical impression described in Prechtl's GM assessment manual [3].

In the present investigation, abnormal fidgety movements were not shown to be an early marker of complex MND at puberty. However, abnormal fidgety movements at 3 to 5 months of age were associated with fine manipulative disabilities at puberty. As fine manipulative disabilities were reported to be significantly related to cognitive difficulties [24], it would have been interesting to determine whether our findings had an impact on academic achievement. However, our cohort attended different types of schools, which rendered an objective comparison impossible.

After Prechtl had discovered the existence of fidgety movements as an age-specific, distinct form of GMs, he speculated about the potential biological function of this transient movement pattern [4,25]. It might be conjectured that one of the ontogenetic adaptive functions of these small movements is optimal calibration of the proprioceptive system. This is supported by the fact that fidgety movements emerge at the 3-month transformation of many neural functions [25], and precede visual hand regard, the onset of intentional reaching, and visually controlled manipulation of objects. As many aspects of the adaptation to the extra-uterine condition are not achieved before the 3rd month post-term, the proprioceptive system is still tuned to the intra-uterine condition. A re-calibration of this sensory system is required in order to achieve proper control of subsequent fine motor activity [3,25]. Similar observations were made in blind infants. Their fidgety movements were exaggerated in amplitude and jerky in character, and their presence lasted longer than in infants with vision. In fact, they were observed until the post-term age of 8 to 10 months [26]. In the latter study the authors speculated that exaggerated fidgety movements might be indicative of an attempt to compensate poor integration of proprioception and vision.

The present study showed that abnormal fidgety movements were only related to a dysfunction of fine manipulative ability at puberty. Particularly visual-motor control, i.e. the ability to coordinate eye and hand was impaired. Our findings do not contradict the conjectured ontogenetic function of normal and abnormal fidgety movements. Rather, they justify further investigation of this subject, which might well have significant practical implications.

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