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Obstetric brachial plexus lesions and central developmental disability☆,☆☆

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ABSTRACT

Aims: First, to assess whether children with an Obstetric Brachial Plexus Lesion (OBPL) have a higher incidence of Central Developmental Disability (CDD) compared to the general population. Second, to test the ability of General Movements (GMs) to identify CDD children already at three months of age.
Study design: A prospective cohort study for infants referred to our tertiary nerve lesion clinic.
Subjects: A prospective cohort study of 38 infants with OBPL followed until 5 years (mean age).
Outcome: Measures quality of fidgety GMs at 3 months; presence or absence of CDD at a mean age of 5 years; severity of the brachial plexus lesion.
Results: Five patients (13%) had CDD: one patient had a cerebral palsy and four showed definite other motor and/or mental problems. There was no correlation between the quality of the GMs at three months and CDD. There was no correlation between the severity of the nerve lesion and CDD. We found a correlation between quality of the GMs and severity of the nerve lesion.
Conclusion: Children with OBPL have a high incidence of CDD. In our cohort fidgety GMs had no predictive value for CDD at a later age.

1. Introduction

Obstetric brachial plexus lesions (OBPL) are caused by traction during delivery [1,2]. The resulting nerve injury may vary from neurapraxia or axonotmesis to neurotmesis and avulsion of rootlets from the spinal cord. The degree of spontaneous recovery correlates inversely with the severity of the nerve lesion.

The upper part of the brachial plexus is most commonly affected, resulting in paresis of shoulder abduction, external rotation and elbow flexion. In more severe cases, the remaining parts of the plexus are also involved.

Fortunately, most children show good spontaneous recovery. Despite important flaws in methodology of natural history studies [3], it may be concluded that the percentage of children with residual deficits probably ranges between 20 and 30% [3].

OBPL is a disorder of the peripheral nervous system. In our tertiary OBPL referral centre, we are regularly confronted with patients with coinciding developmental problems or neurological disorders of the central nervous system. For instance, we saw OBPL children with cerebral palsy (CP), developmental coordination disorder or mental retardation, but also behavioural problems, such as Attention Deficit Hyperactivity Disorder (ADHD). For the purpose of this study, all of these entities are grouped and referred to as Central Developmental Disability (CDD). It appeared to us that CDD occurs more frequently in children with OBPL than in the normal population; to our knowledge, however, systematic research on the presence of coinciding CDD in OBPL patients has not been published so far except for one publication that described a high incidence of developmental and behavioural problems in surgically treated OBPL infants [4].

A correlation between the occurrence of OBPL and CDD seems logical. Children with OBPL usually have a history of a difficult delivery. Such a frustrated delivery is correlated with a higher incidence of performing multiple obstetric manoeuvres [5] and prolonged second stage of labour [6]. An additional consequence of a traumatic delivery could potentially be damage to the central nervous system, which in turn might lead to CDD.

CDD can be predicted by scoring General Movements (GMs) in children of 3 months of age. These GMs are spontaneous movements of all body parts. GMs have shown to be a reliable tool for the prediction of central neurological problems; especially the quality of fidgety GMs at the age of three months has a high predictive value for developmental outcome at later age [7]. Abnormal GM's have an incidence of 5% in the general population [8]. The incidence in the Dutch population of developmental problems is around 4% [9], of cerebral palsy 0.1–0.2%, [10] and of ADHD 3–5% [11].

We studied whether our population of children with OBPL have a higher incidence of CDD as compared to the general population. In addition, we examined whether the GM score can predict which

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OBPL children are at risk of CDD. Finally, we looked at correlations between the severity of the nerve lesion, the quality of the fidgety GMs and the presence of CDD.

2. Method

The present study was part of a larger project on the value of early electromyography (EMG) to predict outcome in OBPL [12]. The study protocol was approved by the Medical Ethics Committee of the LUMC. Patients were actively and prospectively recruited by announcing the study to appropriate medical specialists in The Netherlands. After informed consent, three visits were scheduled around 1 week, 1 month and 3 months of age. Infants who could not be seen before 2 months of age were excluded. In total 48 infants were included in the initial study [12]. The infants were filmed at the age of 3 months (mean 87 days, median 87, range 29) in a supine position during 10 min without interruption. The recordings were independently scored according to a standardized protocol [9] by two experienced investigators (SH and RSvW). The fidgety GMs are usually scored in all limbs; in this study the paretic arm was not included. The complexity, fluency and variety of fidgety movements were classified as normal-optimal, normal-suboptimal, mildly abnormal and definitely abnormal (Table 1). Fidgety GMs can only be reliably scored in the appropriate behavioural states (not crying, awake) [7]. Whenever the patient was not in the appropriate behavioural state the GMs were not scored.

Demographic data and severity of the brachial plexus lesion were additionally scored. The severity of the OBPL was qualified as either severe or mild [12]. A severe nerve lesion was defined as neurotmesis or avulsion of roots C5 and C6 (irrespective of the function of roots C7–C8–T1). A mild lesion was defined as an axonotmetic lesion of C5 and C6 with spontaneous recovery after two years of follow-up manifested by a full range of active elbow flexion, a normal or subtotal range of supination and a normal or nearly normal shoulder function without prominent secondary abnormalities.

Central developmental disability (CDD) was defined in the current analysis as any mental and/or neurological impairment, which was diagnosed or confirmed by an independent specialist, such as the referring paediatric neurologist or paediatric rehabilitation specialist. The final chart review for CDD took place when the infants had a mean age of 4.8 years (range 4.1–5.6).

The statistical analysis was performed with the SPSS statistical package 17 (© SPSS Inc). For correlation of the GMs and the severity of the nerve lesion or presence of developmental impairment we applied the chi-square test and the Mann–Whitney test. A p-value of <0.05 was considered statistically significant.

3. Results

Of the initial 48 infants, two parents decided to abort the study before the third month, because good spontaneous recovery occurred before that time. A video film could not be made due to technical problems in three infants and it was not possible to score the GMs due to an inappropriate behavioural state of five patients. Eventually, 38 recordings could be scored and were further analyzed (Fig. 1). The results of the analysis of the video recordings are presented in Table 2, the inter-observer agreement was 100% (kappa = 1).

Table 1
Classification of quality of general movements [7].

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

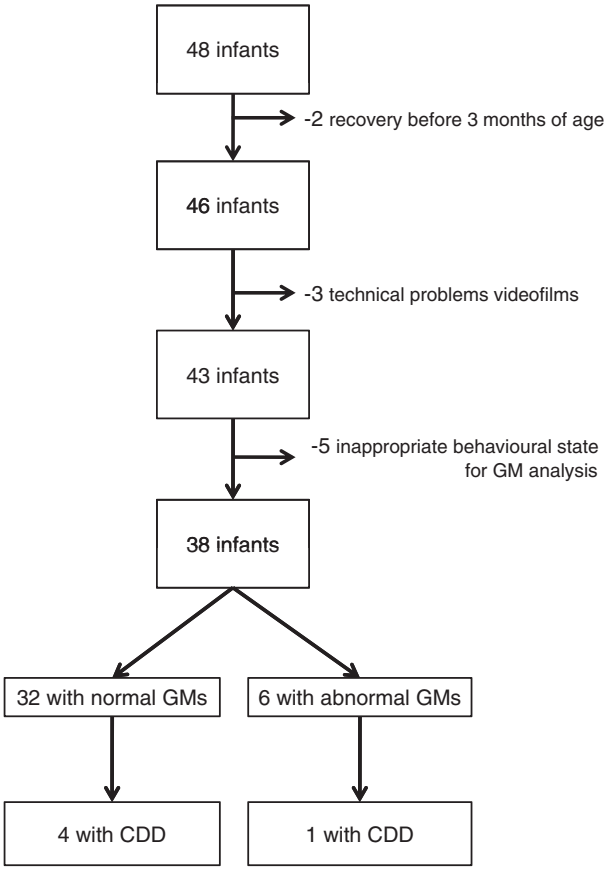

Fig. 1. Inclusion and results.

A total of five patients had CDD during follow-up. One patient had a cerebral palsy, and four showed definite other motor or mental problems. The percentage of patients with CDD was 13% (95% confidence interval 2.4–24%) (Table 3). The patient with a cerebral palsy showed normal GMs. The other four showed abnormal GMs in one patient and normal GMs in three patients (Fig. 1, Table 3).

There was no correlation between the quality of fidgety GMs and the presence of CDD.

Fourteen children had a severe nerve lesion and the remaining 24 children had a mild nerve lesion. In the group of infants with a severe nerve lesion, four infants had mildly abnormal GMs and one had definitely abnormal GMs (Table 2). Of the five patients with CDD, two had a severe nerve lesion and three a mild nerve lesion.

A statistically significant different distribution was found for GM quality and nerve lesion severity (chi-square = 11.790, with 3 degrees of freedom, p = 0.008). Infants with a severe lesion had worse GM scores (Fig. 2). The Mann–Whitney test confirmed statistical significance between the two groups (p = 0.001, two-tailed test).

There was no statistical difference for distribution of CDD and nerve lesion severity.

Table 2
Fidgety GMs, classified by nerve lesion (n = 38).

Fidgety GMs	Severity of OBPL		Total
	Mild	Severe	
Normal-optimal	18	3	21
Normal-suboptimal	5	6	11
Mildly abnormal	1	4	5
Definitely abnormal		1	1
Total	24	14	38

Table 3
Details of five patients with CDD. o = optimal, so = suboptimal, a = abnormal.

Condition	GMs score	Nerve lesion
CP: spastic paresis of both legs	o	Severe
Suspect for developmental coordination disorder and currently examined for epilepsy	so	Mild
Generalised hypotonia and concentration difficulties	a	Severe
ADHD, motor clumsiness	o	Mild
Clumsy motor behaviour, developmental coordination disorder	o	Mild

4. Discussion

The first aim of our study was to investigate the incidence of CDD in OBPL infants (Fig. 3). We found that 5 of 38 (13%) of the study group had a motor or developmental impairment in some way. The exact percentage may, however, not be accurate (95% confidence interval 2.4–24%) as our sample population was small. This is higher as compared to the general population: in The Netherlands, CP has an incidence of 0.15% [10], and CDD/ADHD has an incidence of 5% [13].

It remains speculative how to explain this apparent higher incidence of coinciding CDD in case of OBPL. One hypothesis is that shoulder dystocia is a risk factor for OBPL. During shoulder dystocia, the risk of hypoxic ischaemic encephalopathy increases with prolonged head-to-body interval [14]. Hypoxic ischaemic encephalopathy may result in cerebral palsy [15]. More recently it was described, that mild or moderate asphyxia is also a risk factor for the development of cognitive/executive dysfunction or memory and/or attention problems, such as we found in our patient cohort [16,17].

Our finding of a high incidence of coinciding CDD in OBPL infants, is in accordance with one previous report. Around half of twenty-three nerve surgically treated OBPL infants scored at or above the cut-off score of twelve of the Pre-School Behaviour Checklist [4]. The effects in this cohort were independent of the general condition of the child at birth, as indicated by their Apgar scores.

Our second aim was to test correlations between CDD, quality of GMs and severity of the nerve lesion. In the general population abnormal GM's have an incidence of 4% [8]. In our population, we found that 6 of the 38 children (16%) had a diminished quality of fidgety GMs (5 mildly abnormal, 1 definitely abnormal). This high prevalence

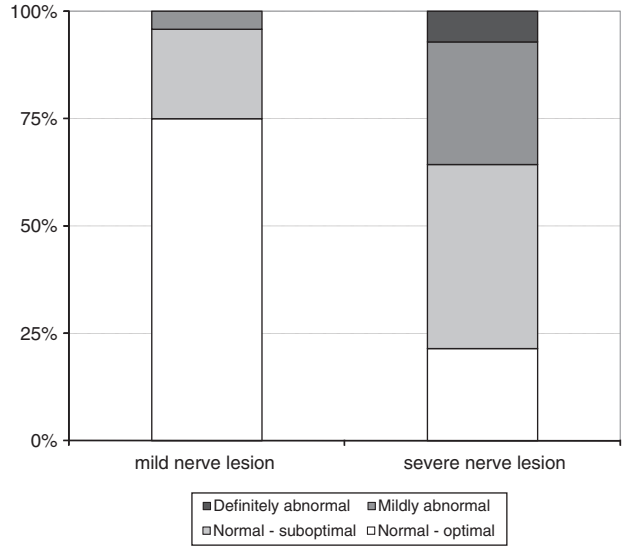

Fig. 2. Stacked column chart showing the distribution of the quality of GMs in severe (n = 24) and mild nerve lesions (n = 14). p = 0.008 (chi-square = 11.790, with 3 degrees of freedom).

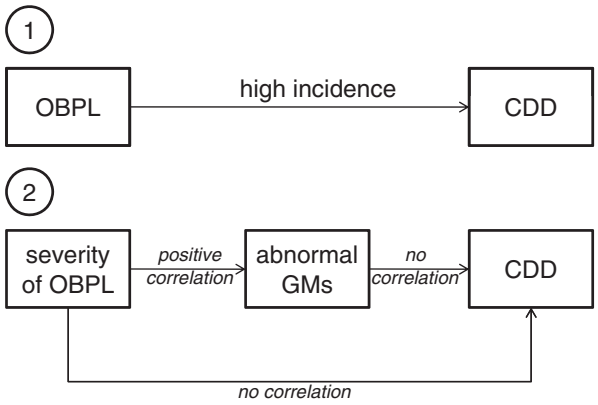

Fig. 3. Results: relationships between OBPL, abnormal GMs and CDD.

could be indicative for a higher risk of these 6 patients for developmental disability at a later age. At follow-up the diminished quality of the GMs could not be correlated to the presence of CDD. There was, however, a correlation between the severity of the nerve lesion and the quality of the GMs: in patients with a severe nerve lesion, an increase in abnormal and suboptimal GMs was found (Fig. 3).

There was no correlation between the severity of the nerve lesion and the presence of CDD. In an earlier study, we studied OBPL patients and the age of independent walking [18]. We found that 80% of children could walk independently at a normal age of 12.1 months (SD 1.8 months, P1: 8.2 months, P99 17.6 months) [19]. There seemed to be a delay in about 20% of children, which did not correlate with the severity of the nerve lesion.

We tried to predict the occurrence of CDD at a later age using GMs at the age of three months. GMs are spontaneous movements of all body parts which can be observed during pregnancy and early childhood. The quality of the fidgety GMs at the age of three months has a high predictive value for development disorders at a later age. Especially the presence of *definitely abnormal* GMs at fidgety age, which implies a total absence of the elegant, dancing complexity of fidgety movements, has a claimed accuracy of predicting cerebral palsy of 85% to 98% in high-risk populations [7]. The remaining 2–15% of infants with definitely abnormal GMs show other developmental problems, such as minor neurological dysfunction, attention deficit hyperactivity disorder or cognitive problems. *Mildly abnormal* GMs at fidgety age are related to the development of minor neurological dysfunction, attention deficit hyperactivity disorder and aggressive behaviour, but the accuracy to predict these problems is modest due to relatively many false positives, resulting in a small positive predictive value [7,20].

In our present patient series only one child was scored as having definitely abnormal GMs. This patient developed a generalised hypotonia and concentration problems. The remaining four children that showed CDD during follow-up scored normal or suboptimal GMs. From the one patient that proved to develop a spastic paresis of both legs, the fidgety GMs were scored as normal.

The reasons why prediction of CDD on the basis of the GMs in our patient group was not possible, remain speculative. As we found CDD in 13% of our cohort, OBPL infants may be considered a high-risk group. Two hypotheses will be discussed.

First, fidgety GMs may not be reliably scored in the presence of an OBPL, due to the presence of upper limb paresis on one side. The diminished quality of fidgety GMs might be the result of the paresis. However, 3-month-old infants were investigated with one or more weighted limbs [21]. Weighting can be considered to result in a movement pattern comparable to that in OBPL. It was found that weighting did not influence the quality of fidgety GMs [21]. The evaluation of GMs in children with an OBPL should therefore, theoretically, not be hindered by the presence of the mono-paresis. Alternatively, the

influence of prolonged weakness in the arm and brain plasticity might influence the GMs in one way or another.

A second reason for the present results could be that the applied methodology to assess developmental disorders was inappropriate. The mean follow-up time of 4.8 years might have been too short. Minor developmental problems are usually only diagnosed in children beyond the age of eight years. In addition, a validated method to assess motor development in OBPL children does not exist. The available scoring methods, like the Bayley Scales of Infant Development [22] and the Movement Assessment Battery for Children [23], depend on bimanual activity of the examined child. It is intuitive that these scoring methods cannot be applied to infants that have an OBPL.

Weaknesses of our study consist firstly of an inclusion bias. The investigated patients were actively recruited for enrolment in the EMG-study and seen at our tertiary referral centre. This may lead to an overestimation of the part of patient with a severe nerve lesion. However, as the present study has a descriptive nature, this should not influence our results. Secondly, the investigators were not formally blinded to the severity of the nerve lesion. Although the affected arm was not taken into account during the evaluation of the fidgety GMs, both investigators have substantial clinical experience in patients with an OBPL to recognise a severe or mild nerve lesion. This might have influenced the scoring of GMs in one way or another.

5. Conclusion

Children with OBPL have a higher incidence of central neurological or development problems. In a cohort of 38 patients with OBPL we found a diminished quality of fidgety GMs at the age of three months. However, this could not be matched to developmental disorders at a later age. There was no relationship between the severity of the OBPL and later developmental problems. Because the incidence of developmental disorders was quite high, the results of this study support a long-term follow-up of infants with an OBPL, with special attention to developmental disorders. A validated development test for OBPL patients should be developed and applied to children in this age group.

Conflict of interest

None.

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