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General movement trajectories and neurodevelopment at 3 months of age following neonatal surgery

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ABSTRACT

Background: Neonates who undergo major surgery are at risk of neurodevelopmental disability. The General Movements Assessment (GMA) is a valid and reliable method to predict neurodevelopment, however, there are minimal data on the applicability among infants post-surgery.
Aim: To describe GMs trajectories following neonatal surgery.
Study design: Prospective cohort study.
Subjects: 217 infants following major cardiac and non-cardiac neonatal surgery.
Outcome measures: Infants were assessed following surgery at term age (mean 40 weeks, SD 2.3), and at 3 months of age (mean 12 weeks, SD 1.6) using the GMA and the Bayley Scales of Infant and Toddler Development III. GMA videos were independently scored by three advanced trained assessors, two blinded to infant details.
Results: The most common result in the writhing period was 'poor repertoire' (n = 117, 54%), however, 99 (84%) of these infants had normal fidgety movements. For infants with normal writhing (n = 75, 34%), only four had absent fidgety movements. Cramped synchronised movements were seen in 10 infants, and three of these were rated as absent fidgety. There was no significant difference between the surgical groups. In total, 24 infants (11%) had absent fidgety movements and lower scores on average in all subtests of the BSID-III than those with normal fidgety movements.
Conclusions: This is the first report describing GMs trajectories in infants who have undergone neonatal surgery. Similar to other high risk infant populations, this group showed a high proportion of poor repertoire writhing movements, however, most infants demonstrated normal fidgety movements and development at 3 months of age.

What this paper adds:

- Poor repertoire movements are most common following neonatal surgery.
- Following neonatal surgery, the majority of infants demonstrate normal fidgety movements.
- Infants with absent fidgety movements also score below average in developmental assessment at 3 months of age.

1. Introduction

Advances in surgical techniques, neonatal intensive care, and imaging have meant that more infants are now surviving early surgery for cardiac and other congenital birth defects [1]. These include cardiac defects such as coarctation of the aorta, hypoplastic left heart syndrome, pulmonary stenosis or transposition of the great arteries, and non-cardiac defects such as diaphragmatic hernia, oesophageal atresia, or gastroschisis. We know that infants who undergo early surgery for these defects are at risk of poor neurodevelopmental outcomes [2]. A New South Wales population based study of infants who had undergone surgery in the neonatal period, found delays across the domains of cognition, language and motor skills at one year of age [2] when assessed using the Bayley Scales of Infant and Toddler Development III (BSID-III), a validated and commonly used developmental assessment. Most notably, 50% of infants who had cardiac surgery showed a delay in gross motor skills on the BSID-III compared to 20% of controls.

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Furthermore, an increase in the prevalence and severity of developmental disorders is seen as the complexity of congenital heart disease increases [3]. Congenital birth defects are also a risk factor for more serious disorders and have been identified as an independent predictor of cerebral palsy (CP) [4].

Ideally, assessments used in the first few months of life should predict which infants will go on to have poor neurodevelopmental outcomes, in order to commence early intervention in the period of maximal brain growth and greatest neuroplasticity. There is emerging evidence that demonstrates early targeted, goal directed therapy is effective in improving outcomes. A recently published Australian randomised controlled trial, used a motor learning, environmental enrichment intervention, titled "GAME" (Goals - Activity - Motor Enrichment) with infants four to six months of age [5]. The GAME intervention resulted in advanced motor and cognitive outcomes at twelve months of age when compared with standard care. The challenge is to use assessment tools to accurately identify those infants at most risk.

Prechtl's Method on the Qualitative Assessment of General Movements (GMA) is a valid, reliable, non-invasive assessment tool, used as a predictor of neurological outcome [6,7]. The GMA is a video based assessment of an infant's spontaneous movement which is evaluated by trained assessors to determine the quality of movement and risk of later neurodevelopmental problems. Movements are categorised within two age related scoring periods. The *writhing* period, from preterm age up until approximately nine weeks post-term age, and the *fidgety* period, from nine to twenty weeks post-term.

Individual general movements (GMs) trajectories are recommended in order to improve both the accuracy of findings, and the prediction of neurological outcome [6]. We would expect infants with normal writhing movements to also have normal fidgety movements at follow-up, and go on to have a normal outcome. However, infants demonstrating persistently cramped-synchronous movements in the writhing period, followed by the absence of fidgety movements at 3–4 months age, are known to be at high risk of cerebral palsy. The most common abnormal GMs result in the writhing period, 'poor repertoire', is less predictive as it can be followed by normal, abnormal, or absent fidgety movements [6].

The GMA has been shown to have excellent sensitivity and specificity for the prediction of both cerebral palsy and minor neurological dysfunction such as developmental coordination disorder [7–12], particularly with the preterm infant population and those with Hypoxic Ischemic Encephalopathy (HIE). However, infants with congenital malformations requiring surgery have often been excluded from these studies, despite their known risks for adverse outcomes, resulting in a lack of information on the use of the GMA with this population. Furthermore, many infants requiring neonatal surgical procedures are born at term [2] and a large proportion of studies reporting on the GMA have focused on the pre-term population.

Our pilot study of the GMA in a large surgical Neonatal Intensive Care Unit (NICU) reported good clinical utility in the infant surgical population, and universal acceptance by parents [13]. We established that a high proportion of infants who had undergone early surgery demonstrated abnormal general movements in the writhing period, with a predominance of poor repertoire movements. These are movements that are monotonous and lack the fluency, variability and complexity seen in normal writhing. It is important to monitor infants with poor repertoire general movements as infants may continue to demonstrate concerning general movements in the 'fidgety' stage, (from approx. 9 weeks post-term age), and go on to have poor neurodevelopmental outcomes. We also demonstrated that there was no significant difference between the GMA results of the cardiac surgical and non-cardiac surgical groups in the writhing period. What we do not know is how these infants are developing at three months of age and what proportion continue to display abnormal movements at follow-up in the 'fidgety' period, predicting risk. Furthermore, it is not yet known whether differences will emerge between the cardiac surgical and non-cardiac surgical groups, placing one group at higher risk of poor neurodevelopmental outcomes.

The aim of this study was to describe the GMs trajectory from the writhing to the fidgety period in a large cohort of infants who had undergone either cardiac or non-cardiac surgery. We also aimed to report on development as assessed on the BSID-III at three months of age, and any differences in the GMs profiles between the cardiac surgical and non-cardiac surgical infants.

2. Method

2.1. Design

This cross-sectional study forms part of a prospective cohort study investigating the use of the GMA to predict developmental outcomes in infants who have undergone major surgery in the neonatal period for either cardiac or non-cardiac related conditions. For the non-cardiac surgery group, major surgery was defined as surgery requiring the opening of a body cavity, such as a laparotomy or thoracotomy. For the cardiac surgery group, major surgery included infants undergoing both open and closed cardiac surgery, but excluded infants who only had patent ductus arteriosus ligation. Ethics approval for the study was obtained through The Sydney Children's Hospital Network, Human Research Ethics Committee (LNR/12/SCHN/494).

2.2. Participants

Infants were eligible if they required major surgery within the first 90 days of life, and met criteria for follow-up in the neonatal developmental follow-up clinic. The clinic follows all infants with congenital cardiac conditions, major surgical anomalies or significant neurological problems. From the cohort of 304 infants, we excluded infants with missing data for the GMA in the writhing or fidgety period. There were 84 infants excluded due to missing data in the newborn period for a writhing GMA. The primary reason was staff availability at the optimum time for assessment. Staff days of work often coincided with infants being unavailable due to theatre or other tests, followed by discharge over the weekend. We also excluded infants who did not proceed to surgery, or who received late complex genetic diagnoses, leaving 217 infants eligible for inclusion in this study (see Fig. 1). These infants had undergone either cardiac surgery (n = 106, 49%), non-cardiac surgery (n = 104, 48%), or both types of surgery (n = 7) (refer to Table 1). Infants were predominantly born at term age (81%) in the normal weight range. There were no significant differences between the surgical groups in relation to gestational age, birth weight or age at assessment. Additionally, there were no significant differences for any epidemiological variables between the full cohort of 304 infants and the study sample of 217 infants.

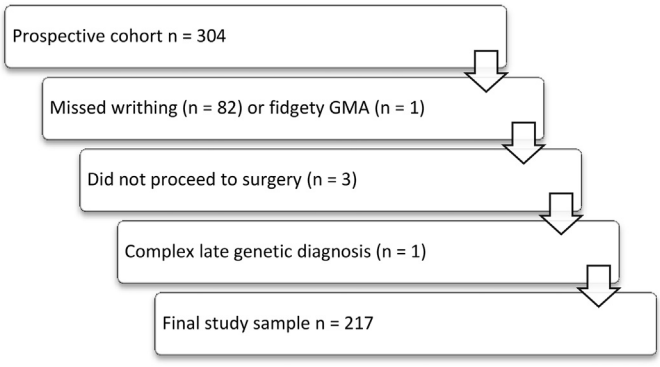

Fig. 1. Study sample.