Developmental Disability at School Age and Difficulty Obtaining Follow-up Data

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abstract

BACKGROUND: The relationship of developmental disability rates with difficulty obtaining follow-up data is unclear. With this study, we aimed to determine if children who attended research follow-up assessments with more difficulty had more disability at school age, compared with those who attended with less difficulty, and to establish the relationship between follow-up and disability rates.

METHODS: Two groups, comprising 219 consecutive survivors born at <28 weeks' gestation or at <1000 g birth weight in the state of Victoria, Australia, in 2005, and 218 term-born, normal birth weight controls were assessed at 8 years of age for neurodevelopmental disability (any of IQ <−1 SD, cerebral palsy, blindness, or deafness). Children were classified as either more or less difficult to get to attend by research nurses involved in the study.

RESULTS: The follow-up rate was 87% for both groups. Overall, children who attended with more difficulty had higher rates of neurodevelopmental disability (42%; 19 of 45) than those who attended with less difficulty (20%; 66 of 328) (odds ratio: 3.09, 95% confidence interval: 1.58 to 6.01; $P = .001$). As the follow-up rate rose among the 3 individual hospitals involved in the assessments, so did the rate of neurodevelopmental disability ($P = .025$).

CONCLUSIONS: Children who attend with more difficulty have higher rates of neurodevelopmental disability at school age than those who attend with less difficulty, and disability rates rise with higher follow-up rates. Rates of neurodevelopmental disability will be underestimated if researchers are not persistent enough to obtain high follow-up rates.

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WHAT'S KNOWN ON THIS SUBJECT: There is disagreement on whether children who are more difficult to get to attend for research follow-up have more neurodevelopmental disability than those easier to get to attend. The relationship between follow-up rates and disability rates is also uncertain.

WHAT THIS STUDY ADDS: The authors of research studies will underestimate rates of neurodevelopmental disability if they do not persist in tracking children who are difficult to get to attend the assessment and achieve high follow-up rates.

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The authors of many cohort studies and randomized controlled trials who have recruited participants in the perinatal period report long-term outcomes for surviving infants in later childhood. Specific research questions are answered for specific topics, such as rates of neurodevelopmental disabilities compared between groups of interest, which might be preterm survivors compared with controls in a cohort study or might be those receiving compared with those not receiving an active treatment in a randomized controlled trial. The information obtained can then be translated into clinical practice, but the data obtained must be accurate.

One problem with long-term follow-up studies is retaining the original participants in sufficient numbers to obtain unbiased answers to the questions being asked. The higher the proportion of participants whose long-term outcomes cannot be determined, the less confidence one has in the findings based just on those able to be assessed. Moreover, the results reported may be biased because of differential dropout rates between the groups of interest.

Recently, Guillén et al[1] systematically reviewed the relationship between follow-up rates and rates of major neurodevelopmental disability in survivors born either at <28 weeks' gestation or at <1000 g birth weight at 18 to 24 months of age. They reported that the lower the follow-up rate, the higher was the rate of disability; for every 1% decrease in the follow-up rate, the rate of disability rose by 0.86%. Conversely, others have reported that preterm children who attended with more difficulty, compared with those who attended with less difficulty, have higher rates of disability, usually because they have lower developmental scores or IQs.[2–5] What is also unclear is the relationship between follow-up rates and outcomes in term-born control infants, because it is often against the outcome of controls that the preterm survivors are being compared. Moreover, being able to predict who is going to be more difficult to get to attend is useful because resources can be directed to establishing multiple contacts and nurturing the relationships with families involved to facilitate their attendance when required.

The aims of this study were to determine (1) if children who attended for research follow-up assessments with more difficulty differed systematically from those who attended with less difficulty with respect to perinatal and sociodemographic variables and neurodevelopmental disability at school age, (2) if there were any differences in these relationships between preterm children and term-born controls, and (3) the relationship between follow-up rates and disability rates. It was hypothesized that children who attended with more difficulty would have more neurodevelopmental disability, that they would be identifiable from sociodemographic data, and that disability rates would rise with higher follow-up rates.

METHODS

Setting and Participants

There are 4 NICUs in the state of Victoria, Australia; 3 are based in high-risk maternity hospitals and 1 is based in a standalone children's hospital. The neonatal units have collaborated with government data collection agencies and the statewide transport service since the late 1970s to obtain population-based data on long-term outcomes for discrete cohorts of the smallest and most immature survivors in the state. Participants for the current study were enrolled in a prospective longitudinal cohort study comprising all 219 consecutive surviving children born either at <28 weeks' gestation or at <1000 g in the state of Victoria in the calendar year of 2005, along with 218 controls who were born at term (37–42 weeks) and with normal birth weight (>2499 g). The sample size for the study was determined by the number of survivors in the 12-month period, which was matched with an equal number of controls. Controls were born in each of the 3 high-risk maternity health care services and were matched for sex of the infant and the mother's country of origin (English the primary language or not) and private health insurance status, as a proxy for social class. Controls were randomly selected from births around the date of the expected date of delivery of the preterm survivors.

Outcome Assessments

Extensive perinatal data had been collected, and the children had been assessed at 2 and 7 to 8 years of age, corrected for prematurity to avoid a known bias in psychological test scores.[6] Results at 2 years[7] and 7 to 8[8] years have been reported elsewhere. At the 7- to 8-year assessment children were assessed by pediatricians and psychologists who were unaware of the child's previous history. Cerebral palsy was diagnosed in children with abnormal tone and loss of motor function, and its severity was determined by the Gross Motor Function Classification System.[9] Blindness was defined as having visual acuity <20/200 in the better eye, and deafness was defined as having a hearing impairment requiring amplification or a cochlear implant, or worse. Cognitive ability was assessed by using the Differential Ability Scales, Second Edition.[10] To standardize cognitive scores, *z* scores of general intelligence (IQ) were computed relative to the mean score (SD 15) for the controls. Before this calculation, the mean for the controls was weighted to reflect the distribution of maternal education and social class of the preterm

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group. Children too impaired to complete the cognitive tests were assigned an IQ *z* score of −4 SD. Neurodevelopmental disability was defined as any of an IQ <−1 SD, blindness, deafness or cerebral palsy. The 3 maternity hospitals were involved in the follow-up assessments, with each hospital being responsible for assessments of children predominantly cared for in their own hospital in the newborn period.

Classification of Difficulty to Get to Attend

Research nurses arranging the follow-up appointments at 7 to 8 years rated the difficulty they had to get the child to attend the appointment. A child who was easy to locate and attended the first appointment made at a mutually convenient time was considered to be less difficult to get to attend the assessment; valid reasons for cancelling appointments, such as acute illness, were acceptable. The remaining children who finally attended assessments were considered more difficult to get to attend. The more difficult group sometimes had multiple broken appointments for no valid reason, or ultimately needed the developmental team to assess the child at another location (usually at their home or school) because parents were unwilling to bring the child in for assessment.

Socioeconomic Variables

Data were recorded on lower maternal education (<12 years of schooling), multilingual status of the household, lower social class (occupation of the main income earner unskilled or unemployed), and whether the family was not intact, with the biological parents not living together.

Ethical Considerations

The study was approved by the Human Research Ethics Committees at the Royal Women's Hospital, the Mercy Hospital for Women, Monash Medical Centre, and the Royal Children's Hospital, Melbourne. Written informed consent was obtained from the parents of all children.

TABLE 1 Participant Characteristics and Difficulty in Getting to Attend Follow-up

Characteristics	Preterm	Term
Survived to 8 y	*n* = 219	*n* = 218
Emigrated, *n* (%)	3 (1)	3 (1)
Lost to follow-up, *n* (%)	9 (4)	4 (2)
Declined follow-up, *n* (%)	15 (7)	22 (10)
Questionnaire only, *n* (%)	2[a] (1)	0 (0)
Assessed at 8 y, *n* (%)	190 (87)	189 (87)
No data on difficulty to get to attend, *n* (%)	4 (2)	2 (1)
Complete data, *n* (%)	186 (85)	187 (86)
Difficult to get to attend, *n* (% with complete data)	24 (13)	21 (11)

[a] One child living remotely was assessed by a pediatrician and free of impairment, but there was no psychological testing.

Statistical Analyses

Data were analyzed by using Stata version 14.2 (StataCorp, College Station, TX).[11] Group differences were compared by using Generalized Estimating Equations, with robust SEs to allow for clustering of children because of multiple births within a family.[12] When the models would not converge, logistic or linear regression models were used. An interaction term for group was added to regression models to determine if outcomes differed substantially between preterm and term groups. Socioeconomic variables were added to the regression analyses to determine their associations with the various outcomes. Data in ordered categories were contrasted by the Mann–Whitney *U* test.[13] Comparisons are presented primarily as odds ratios (ORs) or mean differences, both with 95% confidence intervals (CIs) and *P* values.

RESULTS

Overall, 87% (379 of 437) of both cohorts were assessed for neurodevelopmental disability at 7 to 8 years of age (Table 1). The degree of difficulty in getting the child to attend was not recorded for 6 participants overall (Table 1). Overall, 12% (45 of 373) of children with follow-up data at 7 to 8 years attended with difficulty, 13% (24 of 186) in the preterm group, and 11% (21 of 187) in the control group (Table 1). Children not assessed at 7 to 8 years were similar to those who were assessed for perinatal variables and disability rates at 2 years, in both the preterm and control groups, except that those not assessed at 8 had a higher rate of neurodevelopmental disability at 2 years and their mothers were younger in the control group only (Supplemental Table 5).

On univariable analysis, there were no substantial differences in the mother's age at the time of the birth of the child in those more difficult to get to attend than those who were not, in either the preterm or control groups (Table 2). Children in the preterm group who were more difficult to get to attend the assessment were more likely to have come from a lower social class and have a mother with lower education, but the evidence for associations with other aspects of social disadvantage in the preterm group and with all aspects of social disadvantage in the control group was weaker (Table 2). Children in the control group with a neurodevelopmental disability at age 2 years were more likely to be in the more difficult to get to attend group (Table 2). Combining the groups, on multivariable analysis, having any disability at 2 years (OR: 2.30, 95% CI: 1.15 to 4.61, *P* = .019) and being

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TABLE 2 Perinatal, Sociodemographic, and 2-Year Characteristics of Children Assessed in Preterm and Term Cohorts, Contrasted Between Those Who Were More Difficult to Get to Attend Assessments and Those Who Were Less Difficult to Get to Attend Assessments

Characteristics	Preterm		Term	
	More Difficult	Less Difficult	More Difficult	Less Difficult
Preterm cohort	*n* = 24	*n* = 162	*n* = 21	*n* = 166
Perinatal variables				
Mother's age at delivery, y, mean (SD)	30.8 (5.7)	31.2 (5.8)	31.9 (5.6)	32.7 (5.7)
Outborn, *n* (%)	3 (12)	19 (12)	—	—
Antenatal corticosteroids, *n* (%)	20 (83)	143 (88)	—	—
Multiple birth, *n* (%)	2 (8)	43 (27)	0 (0)	2 (1)
Cesarean delivery, *n* (%)	19 (79)	104 (64)	7 (33)	50 (30)
Gestational age at birth, wk, mean (SD)	27.0 (2.2)	26.8 (1.9)	40.1 (1.1)	39.8 (1.3)
Birth wt, g, mean (SD)	840 (172)	870 (176)	3638 (548)	3587 (479)
Boys, *n* (%)	10 (42)	71 (44)	8 (38)	77 (46)
Exogenous surfactant, *n* (%)	21 (88)	125 of 158 (79)	—	—
Assisted ventilation, *n* (%)	21 (88)	135 of 160 (84)	—	—
Patent ductus arteriosus, *n* (%)	15 (62)	110 (68)	—	—
Grade 3 or 4 intraventricular hemorrhage, *n* (%)	2 (8)	11 (7)	—	—
Cystic periventricular leukomalacia, *n* (%)	2 (8)	3 of 153 (2)	—	—
Necrotizing enterocolitis, *n* (%)	2 (8)	14 (9)	—	—
Postnatal corticosteroids, *n* (%)	5 (21)	30 of 161 (19)	—	—
In oxygen at 36 wk' postmenstrual age, *n* (%)	11 (46)	78 (48)	—	—
Sociodemographic variables				
Lower maternal education, *n* (%)	15 of 23 (65)	63 of 158 (40)[a]	6 (29)	33 of 165 (20)
Lower social class, *n* (%)	13 (54)	49 (30)[a]	4 (19)	18 (11)
Family not intact, *n* (%)	8 of 23 (35)	49 of 161 (30)	7 (33)	29 of 165 (18)
Multilingual, *n* (%)	7 of 23 (30)	23 of 161 (14)	5 (24)	21 of 165 (13)
2-y outcome				
Major neurodevelopmental disability, *n* (%)	14 (58)	70 of 160 (44)	9 of 19 (47)	23 of 162 (14)[a]

—, not applicable.

[a] $P < .05$; denominators are provided if there are missing data.

in a multilingual family (OR: 2.37, 95% CI: 1.07 to 5.25; $P = .034$) were associated with being in the more difficult to get to attend group, but the associations with lower maternal education (OR: 1.50, 95% CI: 0.75 to 3.00; $P = .25$), being from a family that was not intact (OR: 1.21, 95% CI: 0.51 to 2.83; $P = .66$), lower social class (OR: 1.08, 95% CI: 0.46 to 2.54; $P = .86$), and maternal age (OR per year of age: 1.01, 95% CI: 0.96 to 1.07, $P = .66$) were all negligible.

Overall, the corrected age at the time of assessment was greater for the group who attended with more difficulty compared with those assessed with less difficulty (mean difference: 0.35 years; 95% CI: 0.21 to 0.49; $P < .001$). The age difference was larger in the preterm group than in the control group (Table 3). Overall, children who attended with more difficulty had higher rates of any neurodevelopmental disability at 7 to 8 years of age (42%; 19 of 45) than those who attended with less difficulty (20%; 66 of 328) (OR: 3.09, 95% CI: 1.58 to 6.01; $P = .001$). In a multivariable analysis, there was strong evidence that children were more likely to have a neurodevelopmental disability at 7 to 8 years of age if they came from a family with lower maternal education (OR: 2.40, 95% CI: 1.41 to 4.08; $P = .001$) and if they came from a lower social class family (OR: 1.95, 95% CI: 1.05 to 3.64; $P = .035$), but not if they came from a family that was not intact (OR: 1.38, 95% CI: 0.74 to 2.55; $P = .31$), or from a multilingual household (OR: 0.95, 95% CI: 0.46 to 1.95; $P = .88$). When the sociodemographic variables were added, the OR for children who attended with more difficulty having higher rates of any neurodevelopmental disability was slightly lower than the unadjusted analysis (OR: 2.60, 95% CI: 1.20 to 5.63; $P = .015$).

There was weak evidence for an interaction between group and difficulty of assessment on the outcome of major disability ($P = .07$). Within the preterm group alone, children assessed with more difficulty had weak evidence for higher rates of any neurodevelopmental disability than those assessed with less difficulty (OR: 2.32, 95% CI: 0.95 to 5.66; $P = .065$). Within the control group alone, children assessed with more difficulty had substantially higher rates of any neurodevelopmental disability than those assessed with less difficulty (OR: 6.24, 95% CI: 1.99 to 19.6; $P = .002$).

Overall, the mean IQ *z* score was −0.66 (SD 1.00) for children who attended with more difficulty, which was substantially lower when compared with −0.18 (SD 1.00) for children who attended with less difficulty (mean difference: −0.49, 95% CI: −0.80 to −0.18;

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TABLE 3 Outcomes at 7–8 Years of Preterm and Term Cohorts, Contrasted Between Those Who Attended With More Difficulty and Those Who Attended With Less Difficulty at 7–8 Years

Characteristics	Preterm		Term	
	More Difficulty *n* = 24	Less Difficulty *n* = 162	More Difficulty *n* = 21	Less Difficulty *n* = 166
Age when assessed, y, mean (SD)	8.1 (0.4)	7.6 (0.4)[a]	7.9 (0.5)	7.7 (0.5)
Any neurodevelopmental disability, *n* (%)	13 (54)	56 (35)	6 (29)	10 (6)[b]
IQ *z*-score <−1 SD, *n* (%)	10 of 23 (43)[c]	49 (30)	5 (24)	8 (5)[d]
Cerebral palsy, *n* (%)	5 (21)	16 (10)	0 (0)	2 (1)
Blindness, *n* (%)	0 (0)	0 (0)	0 (0)	0 (0)
Deafness, *n* (%)	2 (8)	5 (3)	1 (5)	0 (0)
IQ *z*-score, mean (SD)	−1.00 (1.05)	−0.57 (1.10)	−0.30 (0.81)	0.20 (0.70)

[a] Mean difference: 0.5 (95% CI: 0.3 to 0.6) $P < .001$.
[b] OR: 6.24 (95% CI: 1.99 to 19.6) $P = .002$.
[c] One child with moderate cerebral palsy did not have a formal psychological assessment.
[d] OR: 6.17 (95% CI: 1.80 to 21.2) $P = .004$.

TABLE 4 Follow-up Rates, Categorization of Difficulty Getting the Child to Attend and Neurodevelopmental Disability Outcome at 7–8 Years by Hospital of Follow-up

	Hospital			Mann–Whitney *U* test
	A	B	C	
Survivors to 7–8 y, *n*	145	186	106	—
Child fully assessed at 7–8 y, *n* (% survivors)	121 (83)	160 (86)	98 (92)	$z = 2.00$, $P = .045$
Difficulty to get to attend not documented	3	2	1	—
More difficult to get to attend, *n* (% with data)	4 of 118 (3)	18 of 158 (11)	23 of 97 (24)	$z = 4.50$, $P < .001$
Neurodevelopmental disability, *n* (% survivors with data)				
Both groups combined	20 of 118 (17)	36 of 158 (23)	29 of 97 (30)	$z = 2.24$, $P = .025$
Preterm only	18 of 57 (32)	30 of 82 (37)	21 of 47 (45)	$z = 1.36$, $P = .18$
Control only	2 of 61 (3)	6 of 76 (8)	8 of 50 (16)	$z = 2.34$, $P = .019$

Hospitals ordered by increasing rate of the child being fully assessed. —, not applicable.

$P = .002$). There was little evidence for an interaction between group and difficulty of assessment on IQ ($P = .79$). Within the preterm group alone, children assessed with more difficulty had weak evidence for lower IQ scores (mean difference: −0.43, 95% CI: −0.88 to 0.03; $P = .065$). Within the control group alone, children assessed with more difficulty had substantially lower IQ scores than those assessed with less difficulty (mean difference: −0.50, 95% CI: −0.83 to −0.18; $P = .003$).

Among the 3 individual hospitals where assessments were performed, as the follow-up rate increased, so too did the rates of being difficult to get to attend and of neurodevelopmental disability (Table 4). The evidence for trends of increasing disability with higher follow-up rates was stronger for controls than for children born preterm.

DISCUSSION

One major finding of our study was that children who attended follow-up assessments for research at school age with more difficulty had more neurodevelopmental disability than those who attended with less difficulty. The major contributor to the higher rates of disability was low IQ scores, which were almost 0.5 SD lower in those who were difficult to get to attend. A novel finding of our study was that the association between difficulty in getting the child to attend and more neurodevelopmental disability applied particularly to term-born controls. The age at eventual follow-up was later by ∼4 months in those difficult to get to attend, reflecting the additional time it took to ultimately get the children to attend and complete the assessment. The other major finding from our study is that among the 3 hospitals involved, the follow-up rates, the rates of being difficult to get to attend, and the rates of neurodevelopmental disability were all positively associated, supporting the overall contention that higher follow-up rates are associated with more disability. Of possible sociodemographic variables contributing to the difficulty of attendance, the major reason was that the child lived in a multilingual household. Children with known neurodevelopmental disability in early childhood were also likely to be more difficult to get to attend assessments at school age.

In their systematic review, Guillén et al[1] summarized evidence from other studies, both in adults and children, supporting their contention that lower follow-up rates were associated with higher rates of disability in early childhood, at least in those studies emanating from the United States. Several studies outside the United States involving

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follow-up from the neonatal period have generally reported the contrary, that is, that higher follow-up rates are associated with higher rates of disability and that those more difficult to get to attend have more disability than those who have less difficulty.[2–5] On the other hand, the authors of a study within the United States predicted that toddlers who are compliant with follow-up assessments would have worse developmental outcomes than those are not compliant.[14] Guillén et al[1] acknowledged that different demographic features of participants in US studies compared with studies from outside the United States may help to explain the differences in disability rates between countries. The most likely reason is that the majority of reports of long-term outcomes of extremely preterm or extremely low birth weight infants from the United States come from large teaching hospitals that cater to more disadvantaged families, from which children have more disability, whereas many studies from outside the United States encompass reports from geographic cohorts, where children from all social strata are represented, as is the case in the current study.

It is useful to know in advance who might be more difficult to get to attend so that more resources can be dedicated earlier to ensuring successful follow-up. In a previous study of children of <1500 g birth weight assessed at 5 years of age from 1 of the 3 maternity hospitals involved in the current study, we reported that children of mothers with lower education and without partners at the time of birth were significantly more likely to be difficult to follow-up.[4] Neither of these variables was strongly associated with children being in the attended with more difficulty group in the current study. However, the current study would suggest that resources to ensure high follow-up rates be dedicated to those living in a multilingual household and known to have a neurodevelopmental disability earlier in childhood.

What happens when a child fails to attend a follow-up assessment in clinical practice can vary from what happens in research. In clinical practice, if a child fails to attend, another appointment may be offered, but children who repeatedly do not attend are unlikely to be tracked further. It is usually assumed that the problem for which the child has been managed has resolved, or that the family have decided to go elsewhere. Moreover, resources are limited with regard to the ability to continue to track these families, and there are usually long lists of patients awaiting appointments. Because it is important to have complete outcome data for research purposes to avoid bias in outcome reporting, it is vital to assess as close to 100% of participants as possible. Hence, more resources are required to track families for research and help them to attend the follow-up assessments. The latter can include paying costs for travel or accommodation, or being prepared to send the assessment team to the child's home or school to assess the child. The requirement for additional resources is 1 of the reasons why research follow-up studies are expensive.[15]

It is important to understand that documented long-term outcomes of control children are also affected by follow-up rates in the same direction as high-risk preterm groups. Rather than rely on normative data from standard tests that may not reflect their population, there are many research groups that use control groups to ensure a contemporaneous comparison with peers within the same community. With our results, we suggest that if control group rates of follow-up are even lower than for high-risk groups, then differences between the high-risk group and controls will be exaggerated, given that results from the control group with low rates of follow-up will be an underestimation of the rates of disability. Although there are statistical methods like multiple imputation with which to try to account for missing data, they do not replace the gold standard and accuracy of having the highest follow-up rates possible in research studies.

CONCLUSIONS

Children who attend after more difficulty, regardless of risk group, have higher rates of neurodevelopmental disability at school age than those who attend with less difficulty. Moreover, higher follow-up rates were associated with more neurodevelopmental disability. The authors of studies reporting low follow-up rates will likely underestimate rates of neurodevelopmental disability if they do not persist in encouraging children to attend follow-up assessments. With these findings, we highlight the importance of directing resources into ensuring high follow-up rates in research studies to obtain the most valid results.

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ABBREVIATIONS
CI: confidence interval
OR: odds ratio

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