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Short Communication

Toward Equity in Research Participation: Association of Financial Impact With In-Person Study Participation

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Introduction

Disparities in study participation threaten the generalizability of research findings, hinder innovation, and may compound health inequities for under-represented populations.[1] Representative research participation is a key priority for research and regulatory organizations.[2] Yet, study design can influence the accessibility of research participation. We studied factors related to participation (enrollment and completion of in-person follow-up) in a prospective multicenter observational study of neurodevelopment after neonatal seizures (NCT04337697).

Methods

With parent partners and advocacy organization representatives, we designed a study to evaluate developmental trajectories for children who survived acute provoked neonatal seizures.

Author contributions: Drs. Shellhaas and Glass have full access to all the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis. Drs. Glass and Shellhaas are the co-primary investigators of the listed PCORI contract and National Institutes of Health grant. Shellhaas, Lemmon, Gosselin, Franck, and Glass contributed to concept and design. Glass, Soul, Chang, Wusthoff, Chu, Massey, Abend, Lemmon, Thomas, Numis, Sturza, McNamara, Rogers, McCulloch, and Shellhaas contributed to acquisition, analysis, or interpretation of data. Shellhaas, Lemmon, Franck, and Glass drafted the manuscript. All authors contributed to critical revision of the manuscript for important intellectual content. Sturza performed statistical analysis. Shellhaas and Glass obtained funding.

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Eligible families from nine US centers had completed validated telephone and online measures of parent well-being and child development through age 24 months.[3] When enrolling for childhood follow-up, parents were informed that the protocol included annual remote follow-up and a single in-person neurodevelopmental assessment at age five years. Parents received a $50 token of appreciation and up to $250 to offset travel costs. Families who did not complete in-person testing were encouraged to continue telephone and online survey follow-up. As interim quality measures, we evaluated (1) factors related to childhood follow-up enrollment, including demographic, clinical, and parent/family well-being measures; and (2) whether Impact on Family[4] score, measured when the child reached age five years, was associated with in-person participation.

Results

Among 303 infants enrolled in our original study, 167 enrolled in the childhood follow-up protocol. Enrolled families were more likely to be privately insured, report the child's race as white, and have higher maternal education than families who did not enroll. Otherwise, demographic, clinical profiles, and parent well-being measures at age 24 months did not predict consent to participate in school-age follow-up (Table 1).

Among 94 families who have reached the eligibility for follow-up at age five years, 78 families (83%) completed demographic and parent well-being measures at five years and 64 (68%) presented for in-person testing.

Overall and financial Impact on Family[4] scores were higher (worse) for participants who did not attend in-person testing compared with participants who completed in-person testing (Table 2). These results persisted after adjusting for child insurance type, child race, child ethnicity, and maternal education.

Discussion

In this multicenter study, personal, social, and financial impact of a child's illness on the family most strongly influenced in-person clinical research participation. These findings highlight how structural barriers to participation can threaten equity, accrual, and generalizability of research findings.

Studies relying on neurodevelopmental outcomes have considered in-person testing with standardized neurodevelopmental measures and trained assessors as the gold standard. Our findings suggest that this approach may result in under-representation of

TABLE 1.
Demographic, Clinical, and Parent Well-Being Measures Were Not Associated With Consent to Participate in Long-Term Follow-Up

Variable	Total N = 303	Enrolled in School-Age Follow-up 167 (55%)	Not Enrolled in School-Age Follow-up 136 (45%)	*P* Value
Child and parent demographics				
Child sex (male)	170 (56%)	87 (52%)	83 (61%)	0.12
Child race				
American Indian/Alaska Native	2 (0.7%)	1 (0.6%)	1 (0.7%)	
Asian	20 (6.6%)	10 (6%)	10 (7.4%)	
Black/African American	36 (12%)	15 (9%)	21 (15.4%)	
White	192 (63%)	120 (72%)	72 (52.9%)	<0.0001
Native Hawaiian/other Pacific Islander	2 (0.7%)	0 (0%)	2 (1.5%)	
Other	25 (8.2%)	9 (5.4%)	16 (11.8%)	
Unknown/not reported	26 (8.6%)	12 (7.2%)	14 (10.3%)	
Child ethnicity				
Hispanic or Latino	47 (16%)	19 (11%)	28 (21%)	0.12
Not Hispanic or Latino	246 (81%)	141 (84)	105 (77%)	
Unknown/not reported	10 (3.3%)	7 (4.2%)	3 (2.2%)	
Child health insurance				
Public	128 (42%)	56 (34%)	72 (53%)	0.0005
Private	174 (57%)	111 (67%)	63 (46%)	
Unknown	1 (0.3%)	0 (0.0%)	1 (0.7%)	
Maternal education				
More than high school	221 (73%)	143 (86%)	78 (57%)	<0.0001
High school or less	68 (22%)	21 (13%)	47 (35%)	
Unknown/not reported	14 (4.6%)	3 (1.8%)	11 (8.1%)	
Clinical outcomes at age 24 months				
Cerebral palsy, No. (%)	80/273 (29.3%)	47/165 (28.5%)	33/1108 (30.6%)	0.71
Epilepsy, No. (%)	37/282 (13.1%)	21/166 (12.6%)	16/116 (13.8%)	0.78
WIDEA-FS score*	151.9 (33.2)	153.0 (31.8)	150.1 (35.5)	0.48
Family well-being: measured at age 24 months [mean (S.D.)]				
HADS depression	3.3 (3.1)	3.4 (3.1)	3.2 (3.2)	0.67
HADS anxiety	6.0 (4.1)	6.0 (4.1)	6.1 (4.3)	0.86
24 m WHO overall QOL	76.6 (18.2)	77.2 (17.6)	75.5 (19.3)	0.50
24 m IES total	14.9 (14.4)	14.9 (14.4)	14.8 (14.4)	0.92
IOF Overall impact	28.0 (10.3)	28.4 (10.2)	27.3 (10.6)	0.43
IOF financial impact scale	9.2 (3.4)	9.2 (3.4)	9.3 (3.6)	0.98
IOF coping scale	9.0 (2.8)	9.0 (2.7)	8.9 (3.1)	0.77

Abbreviations:
HADS = Hospital Anxiety & Depression Scale
IES = Impact of Events Scale (measures symptoms of post-traumatic stress)
IOF = Impact on Family (measures impact of caring for a medically complex child)
WHO = World Health Organization
WIDEA-FS = Warner Initial Developmental Evaluation of Adaptive and Functional Skills
QOL = Quality of Life
* WIDEA-FS score for typically developing children at this age is 172 ± 10 points.

TABLE 2.
Higher Scores on the Current Impact on Family Scale (at Child Age 5 Years) Were Associated With Lower Participation in In-Person Developmental Testing

	Family Did Not Come to 5-yr Visit	Family Attended 5-yr Visit	*P* Value (Unadjusted)	*P* Value (Adjusted*)
5-year IOF, overall	32.2 (10.9)	24.9 (9.9)	0.005	0.005
5-year IOF, financial	9.7 (3.5)	7.8 (3.5)	0.0025	0.016
5-year IOF, coping	9.0 (2.3)	9.0 (2.8)	0.93	0.93

Abbreviation:
IOF = Impact on Family
The IOF overall score represents a single construct of personal, family, and social impact (15 items; range 15 to 60), with higher scores indicating a greater impact on the family. In addition, two subscales, financial strain (four items; range 4 to 16, higher scores indicate more financial strain) and coping (six items; range 6 to 20, higher scores indicate worse coping) are measured separately, but not included in the overall score.
* Adjusted analyses controlled for child insurance type—public or private, child race, child ethnicity, and parent-reported maternal education level.

key groups—even among families who were interested in research participation. Some parent-reported measures, which can be administered remotely, have concurrent validity with in-person assessment.[5] Future work should prioritize the development and evaluation of outcome measures that can be administered in teleconference or remote settings.

Completing in-person study procedures can result in direct and indirect costs to participants, including lost wages, risk of job loss, transportation, childcare, and elder care. Study budgets and designs must be attuned to these realities. Strategies to mitigate the logistical and financial impact of study participation include increased incentives, flexible appointment scheduling, and remote or in-home data collection.[1] Families may also face less easily quantified challenges to research participation, such as risk for infectious exposure in a medical setting and concerns about missing school or therapies. Decentralized clinical trials and direct-to-family approaches may improve participant retention and enhance diversity.[6] Increasing the racial, ethnic, and cultural diversity among the research team and including parent partners on the study team are strategies to improve the feasibility and acceptability of protocols and increase trust in clinical research and motivation leading to greater participation.

Efforts to improve representation in research participation, diversity in research populations, and rigor in study design call for creative solutions. Parent-reported and teleconference-based assessments may provide cost-effective, valid, and inclusive solutions to this important priority and should be considered for research studies that include neurodevelopmental outcomes.

Data sharing statement

The authors will provide deidentified data to qualified investigators upon reasonable request, after the primary outcomes of the parent study (NCT04337697) have been published.

Declaration of Competing Interest

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