

Participation of young severely disabled children is influenced by their intrinsic impairments and environment

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A prospective study of the families of young severely disabled children applying to a non-governmental organization for financial support examined the respective contributions of a child's intrinsic impairment and the family's environment to their participation. The study population comprised the families of 600 children of median age 2 years 8 months (range 1y–12y 1mo), of whom 46% had autistic or severe behaviour difficulties, 12% other learning disabilities, 6.8% cerebral palsy, and 10% non-neurological causes of severe disability. Intrinsic impairment was measured with the Health Utilities Index (HUI); environmental factors using a novel instrument, the European Child Environment Questionnaire (ECEQ); and participation of child and family using the Lifestyle Assessment Questionnaire. Principal components analysis was used to examine interrelationships. A five-component model explains 50% of the variance in the ECEQ. The first component, ECEQ1, reflects the support a family receives from professional services and family. HUI and ECEQ1 were independently associated with participation ($p < 0.0001$) with comparable effect sizes. We conclude that the participation of young severely disabled children is affected to similar extents by intrinsic impairment and environmental factors such as services, support, and attitudes.

Extremes of the medical and social models of disability exaggerate respectively the contributions of intrinsic impairment and environment to the disadvantage experienced by disabled people.* The International Classification of Functioning, Disability and Health (ICF)² achieves a qualitative synthesis of these models. Participation is defined as involvement of the person in life situations, and replaces the concept of 'handicap'. Participation is recognized to be determined by both intrinsic impairment and environmental or contextual factors. The latter include not just attributes of the physical environment, but also societal values as reflected both at the macro level of public legislation and policy, and the micro level of the attitudes as experienced of friends, family, and public.

There is a need to explore this model quantitatively. What are the relative importance of intrinsic impairment (the target of medical interventions) and contextual factors (the target of political, social, and educational interventions) in determining participation? The ICF model predicts that individuals with similar levels of intrinsic impairment will achieve differing levels of participation in different settings but does not as such predict the magnitude or importance of these effects. In a population-based study of children with cerebral palsy (CP) in the northeast of England, Hammal et al. confirmed that participation was determined in part by the family's district of residence,³ although the factors making a particular district more or less facilitatory were not identified. The present study extends this approach to a national cohort of children with a wide variety of severe impairments. In contrast to the study by Hammal, we recognized that the small number of children living in any one district in this study would preclude demonstration of empiric area-of-residence effects on participation. Rather, we hypothesized that participation would be affected by properties of the experienced environment that could be measured for each child and compared across the cohort.

Method

The Family Fund (FF) is a UK non-governmental organization that gives financial support to low-income families of severely disabled children with a wide variety of medical conditions. Ethical approval for the study was given by the trustees of the FF. During the standard process for the consideration of an application for funding, families applying for the first time were invited to join the study, without prejudice, and on a self-selecting basis. Written information given to families stated that funding decisions would not depend on participation in the research and that assessors of applications would not know whether a family had participated. Parents gave written agreement to join the study. Participating families completed three questionnaires that were distributed by post and collected by an FF worker during their routine visit. Questionnaire data were anonymized by FF staff before being passed to the research group for analysis. The instruments used were the Health Utilities Index 3 (HUI);⁴ the European Child Environment Questionnaire (ECEQ), a novel environmental instrument; and the Lifestyle Assessment Questionnaire (LAQ).⁵

*Indeed, these models drive a usage debate,¹ with arguments from each viewpoint that both 'disabled person' and 'person with disability' reflect enlightened attitudes. The former will be used for consistency in this paper, in sympathy with the social-model perspective that the person is 'disabled by' discriminatory attitudes and inadequate resources. Whilst impairment may be an intrinsic property of the person, disability is not: it is 'done to you'.

See end of paper for list of abbreviations.

The HUI is a measure of intrinsic impairment that aggregates a weighted sum of simple evaluations of vision, hearing, speech, ambulation, dexterity, emotion, and cognition into a unidimensional multi-attribute utility function (HUI-MAUF) score. The weightings used in this study were derived from a consultation exercise involving Canadian adults asked to rate the 'value' of hypothetical combinations of individual domain scores (in adults) on a unidimensional scale.⁶ On the scale, immediate death has a utility of zero, and perfect health a utility of one. Although several paediatric measures of 'disease severity' exist, such as the Pediatric Evaluation of Disability Inventory (PEDI),⁷ the HUI was chosen because of three properties essential to this study: its broad applicability across a range of diagnoses; its derivation of a unidimensional summary score; and, most importantly, its strictly 'within the person' focus. It is emphatically a measure of impairment, i.e. function at the level of body organs such as vision, hearing, and dexterity. Many alternative candidate measures such as the PEDI stray into activity- and participation-level effects of disease.

Table I: Format of typical European Child Environment Questionnaire item

Does your child receive practical physical help from teachers, therapists, and helpers at school?	
A	Not needed or not relevant to my child
B	Mostly yes, and this helps my child a lot in everyday life
C	Mostly yes, and this helps my child a little in everyday life
D	Mostly no, but this only restricts my child a little in everyday life
E	Mostly no, and this restricts my child a lot in everyday life

The ECEQ is a novel instrument developed to capture the environment relevant to disabled children in a European study

Table II: Selected characteristics of study sample

Male, %	70
Age, y:m	Range 1–12:1; median 2:8, IQR 1:6
HUI-MAUF score	Range –0.3 to 1.0; median 0.44, IQR 0.47
Employment status of main carer ^a	21% employed full- or part-time or self-employed
Rented housing	44%
Ethnic origin ^a	85% White British
Uses wheelchair (parent reported)	7.2%
Behaviour problems (parent reported)	45%
Primary condition category	Autism/severe behaviour problems: 46% Learning disability*: 12% Non-neurological disability: 10% (umbrella category for severe respiratory, cardiac, and other non-neurological medical conditions) Cerebral palsy: 6.8% Other disorders of the CNS: 5.5%
Index of Multiple Deprivation (IMD) quintiles (derived from residence postcode; available for England and Wales only)	Quintile 1 (greatest deprivation): 12.7% Quintile 2: 40.7% Quintile 3: 23.4% Quintile 4: 11.0% Quintile 5 (least deprivation): 12.2%

HUI-MAUF, Health Utilities Index Multi-attribute Utility Function; IQR, interquartile range (i.e. range enclosing central 50% of data); CNS, central nervous system; ^aMissing data for more than 25% of individuals.

*North American usage: mental retardation.

Table III: Component loadings for a five-component categorical principal components analysis model of European Child Environment Questionnaire. For each component, up to 20 (of 45) variables are shown in loading order where loading >0.3

	<i>Component 1</i>	<i>Component 2</i>	<i>Component 3</i>
Respite overnight	0.58	Lifts in public	0.74
Few hours break	0.58	Ramps in public	0.69
Home helper	0.57	Smooth pavements in public	0.67
Services coordinate	0.57	Ramps at school	0.66
Emotional support teachers	0.56	Room in public	0.63
Friends positive	0.55	Get equipment grants	0.62
Practical help from friends	0.55	Toilets in public	0.62
Practical help from public	0.53	Adapted toilets at school	0.60
Encouraged by teachers	0.52	Get home modernization grants	0.46
Emotional support from friends	0.52	Adequate parking	0.45
Professionals coordinate	0.52	Lifts at school	0.39
Practical help teachers	0.52	Home adaptations	0.36
Encouraged by classmates	0.51	Accessible taxis	0.36
Professionals listen	0.51	Accessible trains	0.34
Counselling available	0.51	Accessible buses	0.31
Emotional support classmates	0.51		
Teachers understand condition	0.48		
Parent support groups	0.47		
Special staff at school	0.47		
Fully included at school	0.46		
Proposed name	Support	Physical accessibility	Educational provision
Cronbach's alpha	0.90	0.86	0.71
% variance accounted for	18.0	13.4	7.4

of children with CP (SPARCLE, Study of the Participation of Children with Cerebral Palsy Living in Europe⁸). Questions systematically probe areas identified as important to these families through qualitative studies:^{9–11} physical attributes of home, school, and public spaces; provision, appropriateness, and adequacy of services; and the experienced attitudes of wider family, friends, professionals, and the public. Item performance has been evaluated in extensive pilot studies as part of the SPARCLE project (A Colver, personal communication). The standard form of the items establishes firstly, whether a given factor is present or absent, and secondly, its importance to the family (Table I). For this study, responses were treated as categorical (i.e. no particular ordering was assumed) and a form of principal components analysis (PCA) adapted to handle categorical data was used^{12,13} to derive a structure for the ECEQ.

The LAQ was developed to measure the impact of disability on young children and their families across a range of medical diagnoses,⁵ a concept that for this study was regarded as equivalent to participation (see Discussion). It is a generic adaptation of the CP-specific instrument used as a measure of participation in our previous work in this field.³ Six sub-domain scores are generated for: communication, mobility, self-care, domestic life, interpersonal interactions and relationships, and community and social life. The first component of a conventional PCA of the six sub-domain scores was used as a unidimensional overall participation score (LAQ1) as the dependent variable in further analyses (see Discussion).

The child's primary medical condition as recorded by the FF was mapped to one of 11 broad condition categories.

The basic hypothesis of this paper, that participation is

determined by both intrinsic impairment and contextual factors, was examined quantitatively in a conventional multiple linear regression. LAQ1 was the dependent variable, and HUI-MAUF and the five ECEQ components were simultaneously block-entered as independent variables. A significance threshold of $p < 0.05$ was used.

In subsidiary analyses, each of the six LAQ sub-domain scores in turn were entered as dependent variables in multiple regressions with the same independent variables. These were repeated in each of the primary condition category subpopulations. Owing to the multiple comparisons in these analyses, a more stringent significance threshold of $p < 0.001$ was used.

Published government indices of deprivation (measures of education and skills, healthcare provision, employment, and income levels) and a summary Index of Multiple Deprivation (IMD)¹⁴ were obtained for the family's home postcode (not available for Scotland). LAQ sub-domain scores were also regressed against these deprivation scores.

Statistical analyses were performed using SPSS (Version 11) for Apple Macintosh OS X.

Results

The first 600 completed forms were used from 1242 families approached (48% response rate), with no family contributing more than one child. No significant difference was noted between responders and non-responders apart from a slight over-representation of families with children with autistic and behavioural difficulties among responders (data not shown). The severity of the children's impairments is reflected both in the young age at which the families had approached the FF and their low HUI-MAUF scores (Table II).

A primary condition category was known for 594 children (Table II). ECEQ data were available for 589 children. Categorical PCA analysis suggested a five-component model, accounting for 48% of the variance (Cronbach's alpha 0.975). The items in each component, component loadings, and proposed component names are shown in Table III. LAQ data were available for 598 children and the first component of a conventional PCA (designated LAQ1) accounted for 44% of variance in LAQ.

In a multivariate regression model with LAQ1 as dependent variable and the five ECEQ components and HUI-MAUF block-entered as independent variables, 48% of variance in LAQ1 was accounted for (Table IV). HUI-MAUF and three ECEQ components (support, physical accessibility, and transport) were independently statistically significantly associated with LAQ1 ($p < 0.05$). The effect sizes of HUI-MAUF and the first ECEQ component (ECEQ1; 'Support'), as judged by standardized beta, were comparable (Table IV). The standardized betas demonstrate that although the ECEQ physical accessibility and transport variables also showed statistically significant associations with LAQ1, these were quantitatively weaker. The negative beta for HUI-MAUF arises from sign conventions, with high HUI-MAUF scores representing increasing health, but high LAQ scores representing poor participation.

Further regression analysis, using each of the LAQ sub-domains as dependent variables in turn, within each primary condition group allowed more detailed examination of which aspects of participation were being affected by which aspects of the environment. The regressions are too lengthy to report in full but, for example, for children with autistic and behavioural disorders Support (ECEQ component 1) was associated

Table III: continued

Component 4		Component 5	
Accessible buses	0.69	Emotional support home	0.47
Adequate bus service	0.69	Practical help home	0.44
Accessible trains	0.56	Emotional support friends	0.42
Accessible taxis	0.51	Practical help friends	0.40
Adequate car	0.50	Home family positive	0.32
Transport		Family and friends	
0.61		0.43	
5.5		3.9	

only with the LAQ domains reflecting interaction with family and the public. Likewise, although Physical Accessibility of the environment (ECEQ component 2) was associated with the self-care LAQ domain for children with conditions of the central nervous system (who might be expected to have mobility difficulties that make them sensitive to the physical accessibility of their environment), the demonstration of the same effect to a lesser degree for children with autism and behavioural disorders was unexpected.

No significant relation was demonstrable between any of the LAQ sub-domains and any of the postcode-derived deprivation indices.

Discussion

This study provides empirical support for an integrated socio-medical model of disability. The demonstration of similar standardized beta regression coefficients for a measure of intrinsic impairment (HUI-MAUF) and a measure of community support (ECEQ Support) on a measure of children's participation (Table IV) is one of the first quantitative examinations of the magnitudes of 'medical' and 'social' influences on participation in such a population of severely disabled children. Previous similar studies have been confined to single-diagnosis groups. In a study of adults with spinal cord injury, 5% of variance in participation was explained by environmental factors,¹⁵ although the authors only examined the extent to which environmental factors obstructed, rather than facilitated, participation. A small study of children surviving traumatic brain injury¹⁶ suggested 10% of variance in participation was explained by environmental factors.

The subgroup analyses described also show the power of this approach to identify the effects of specific environmental variables (ECEQ components) on particular subdomains of participation within different condition categories in ways that might inform policy. For example this study suggests that improving environmental Physical Accessibility will have little effect on any aspect of the LAQ for children with learning disabilities, but improving Support may benefit aspects of par-

ticipation reflected in the domestic life domain of the LAQ. ECEQ component scores offer a potentially useful new measure of the properties of local areas, particularly as they reflect family-reported perceptions of these factors, and examining variation in these scores between areas may be informative.

A previous study³ showed independent associations between participation and district of residence for children with CP but was unable to identify the district-level factors influencing participation. In the present study, it is Support, Physical accessibility, and Transport (Table IV) that influence participation. Only some are 'macro' level factors that might be influenced by local authority policies and spending priorities. The Support component in particular appears in part to reflect 'micro' level cultural attitudes of family, friends, and the public. This might be one reason for the interesting lack of a demonstrable association between participation and postcode-derived deprivation indices, which was also a finding of our previous study.³ However, there may be other reasons. The FF only accepts applications from families with incomes below a threshold value (approximately £23,000) and, therefore, deprived families and districts are over-represented in this study; associations might be found if more children from less deprived backgrounds had been included. Another explanation may relate to the very high spatial resolution of IMD scores, which are reported for areas with a population of approximately 1500. In the UK, educational and other relevant policy decisions are made at an administrative level covering populations of tens or hundreds of thousands, and demonstration of effects of these policies on participation may require use of socioeconomic indices aggregated at this scale.

The main limitation of this study is the post hoc derivation of the component structure of the ECEQ, which may mean some of the interaction patterns are specific to this population. For example, a surprising lack of association between Physical Accessibility and any LAQ-subdomains in the CP group (not shown) may reflect the young age of this cohort: physical accessibility, adaptation, and mobility issues may be more pertinent to the families of older, heavier children. The applicability of these findings to other populations should be prospectively tested, although we believe the basic finding of independent effects of both impairment and environment on participation will be generalizable. Although the total percentage variance in the ECEQ accounted for in the five-component model is relatively low at 48%, this is in part a consequence of the large number of ECEQ items. The internal validity of the model is supported by the Cronbach's alpha and the face validity of the items loading on each of the components in Table III. The 48% response rate and slight over-representation of families of children with autistic and challenging behaviour is typical of previous FF surveys.

As the final effect of disability on both child and family, measurement of participation has the potential to allow comparisons of the impact of childhood morbidities of all kinds. Measurement of participation is also the only way to evaluate an intervention such as the provision of support services, aimed at mitigating the effects of impairment rather than the impairment per se.¹⁷ However, although attractive for analytical purposes, the search for broadly applicable unidimensional scales of participation may be naïve. Previous validations of unidimensional measures of participation were within single-condition groups (e.g. acquired brain injury¹⁶). The approach taken by the creators of the HUI to aggregate domains

Table IV: Regression of European Child Environment Questionnaire (ECEQ) components and Health Utilities Index (HUI) onto Lifetime Assessment Questionnaire (LAQ1). The sign conventions are such that positive scores reflect health in the HUI, poor participation (severe impact of disability) in the LAQ, and an obstructive, unhelpful environment in the ECEQ

	<i>Standardized coefficients (beta)</i>	<i>Significance (p)</i>
(Constant)		0.000
ECEQ Component 1 (Support)	0.34	0.000
ECEQ Component 2 (Physical accessibility)	0.10	0.002
ECEQ Component 3 (Educational provision)	0.06	0.065
ECEQ Component 4 (Transport)	0.07	0.021
ECEQ Component 5 (Family and Friends)	0.03	0.379
HUI-MAUF	-0.45	0.000

MAUF, Multi-attribute Utility Function.

of impairment into a unidimensional HUI-MAUF score was to ask panels of the public to rate hypothetically the relative desirability of arbitrary combinations of impairments in adults on a 0–1 scale. As such, it is used as a relative, not an absolute, scale of impairment in this study. Any inference that the health status of the 12% of the children in this study with negative HUI-MAUF scores should be regarded as literally 'worse than immediate death' is inappropriate, as the weightings were not established in this population. Comparable approaches were used to create a unidimensional score for the previous version of the LAQ designed specifically for children with CP.^{18,19} Achieving a unidimensional score for the generic version of the LAQ used in this study would require consensus societal views on how to assign relative weightings to, for example, the mobility impairments of a child with severe CP versus the social interaction impairments of a child with severe autism, which is arguably a much greater challenge. The subjective nature of such a comparison was circumvented in this study by the empiric PCA approach.

As typically applied in an adult setting, the concept of participation implicitly regards the individual as autonomous, trying to participate because an activity matters to him or her. In the context of the young child this is less appropriate, and, for this study, participation was considered at the level of the family. As such, the LAQ is a measure of participation of severely disabled children and their families.⁵ This study does, however, highlight limitations of current instruments. The measurement of participation, particularly in children, is still a challenge.²⁰

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List of abbreviations

ECEQ	European Child Environment Questionnaire
FF	Family Fund
HUI	Health Utilities Index
HUI-MAUF	HUI Multi-attribute Utility Function
LAQ	Lifestyle Assessment Questionnaire
PCA	Principal components analysis
PEDI	Pediatric Evaluation of Disability Inventory