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Regional Variation in Survival of People With Cerebral Palsy in the United Kingdom

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ABSTRACT. *Objectives.* **Regional variation in survival of people who have cerebral palsy (CP) has been observed but not previously investigated in detail. In addition to true differences, variations in the methods and definitions used, completeness of ascertainment, and the role of potential confounding factors all have been proposed as possible explanations for these observed variations. Our aim was to assess the regional differences in survival of young people with CP and the effect on survival of socioeconomic differences after adjustment for variations in level of impairment and birth characteristics.**

Methods. **Survival patterns for young people with CP were calculated using information from a collaborative database. This database consisted of registrations of children who were born with CP in 5 geographically defined areas in the United Kingdom between 1980 and 1996. Notification of subsequent deaths was provided by the relevant births and death register. We consider the effects of birth characteristics, socioeconomic status, and severity of CP on survival.**

Results. **There were 325 deaths among the 4007 cases of CP identified. The proportion of affected children who survived to 20 years of age ranged from 85% (Mersey-side and Cheshire, male individuals) to 94% (North of England, male individuals). Multivariate modeling showed that the severity of impairment had the biggest impact on survival and that additional contributions were made by birth weight and socioeconomic status but that after such adjustments regional differences were no longer significant.**

Conclusions. **The number and the severity of impairments are the best predictors of survival in young people with CP. After adjustment for the number of impairments, children who were born in affluent areas and had a low birth weight have an increased risk for death over those who were born with a normal birth weight. The same does not hold, however, for those who were born in deprived areas.** *Pediatrics* 2005;116:1383–1390; *survival, impairments, birth weight, socioeconomic status.*

ABBREVIATIONS. CP, cerebral palsy; UKCP, United Kingdom Collaborative Network of Cerebral Palsy Registers; CI, confidence interval.

Previous work has reported that most children with cerebral palsy (CP) survive to adulthood, unless they have severe motor and cognitive impairment.[1–9] Some variations have been observed in reported survival rates from different regions and countries. However, much has been attributed to differences in severity profiles of the groups studied. Indeed, after conditioning on severity of impairment, country variations between California and Western Australia were no longer evident.[10] In contrast, reported survival rates from England among the group with severe impairments do seem to differ.[7] It is unclear from the published work whether these regional variations observed in England reflect true differences, for example, reflecting regional variations in general survival patterns, risk factor profiles, or levels of care, or rather are artifactual, for example, relating to differences in definition or measurement of impairments or differences in the ascertainment of those with the most severe impairment.

The United Kingdom Collaborative Network of Cerebral Palsy Registers (UKCP) brings together 5 research groups, each looking at the prevalence and the severity of CP among children who were born within separate, defined geographic areas in the United Kingdom. Issues of variations in ascertainment and in the definition and the measurement of impairments have been addressed. It therefore is possible to determine whether regional differences in survival are maintained after adjustments for variations in levels of impairment and birth characteristics. Furthermore, the effect of socioeconomic status on survival into adulthood for children who are severely affected by CP was investigated.

METHODS

UKCP

The 5 participating registers were the Merseyside and Cheshire Cerebral Palsy Register[2,9,11]; the North of England Collaborative Cerebral Palsy Survey[7,12]; the Northern Ireland Cerebral Palsy Register[13]; 4Child—the Four Counties Database of Cerebral Palsy, Vision Loss and Hearing Loss in Children[14,15] (formerly the Oxford Register of Early Childhood Impairments); and the Scottish Register of Children with a Motor Deficit of Central Origin.[14,16] Henceforth, the registers will be referred to as Mersey, North of England, Oxford, Northern Ireland, and Scotland, respectively. The geographic regions covered by these registers were for Mersey the counties of Cheshire and Merseyside; for the North of

England the former health districts of Northumberland, Newcastle, and North Tyneside until 1990 and the former health Northern region after 1990; for Oxford the counties of Buckinghamshire, Berkshire, Northamptonshire, and Oxfordshire; and all of Northern Ireland and Scotland. Full details of assessment, ascertainment, and consent methods for each of the registers may be found in the references.

A collaborative database to which the 5 registers submitted anonymized details of their cases was established. Cases were defined as children with early-impairment CP, that is CP that is believed to be attributable to an insult that occurred before age 28 days. Data on gender, birth weight, gestational age, year of birth, type and levels of impairments, and socioeconomic status were provided.

The 5 registers that contributed to the collaboration were established at different times, and the periods for which cases were available varies by register. Cases were included from year of birth 1980 for Mersey and the North of England and available from 1981 for Northern Ireland and 1984 for Oxford and Scotland. The North of England, Northern Ireland, and Oxford provided information on births up until 1996, whereas Mersey and Scotland had information on births until 1989. Cases from all 5 registers were followed up for death notifications until March 31, 2004. Each register flagged its cases with the relevant births and deaths register (the National Health Services Central Register for England and the Northern Ireland Central Services Agency or General Register Office for Scotland), which notifies the register when a flagged person dies. The U.K. system of death registration, in which by law all deaths must be registered, ensures that the fact of death is recorded for virtually all people. Dates of death for those who are abroad with the armed forces were also recorded, as were dates of emigration. Lifetimes were computed in weeks from birth until death. For those still alive, lifetimes in weeks from birth until the chosen censoring date, March 31, 2004 (or date of emigration), were computed. This censoring date had a lag of ~3 months to allow for any delays in notifications of deaths.

Our inclusion criteria were that cases had to be flagged with the relevant births and deaths register, alive at age 2 years, and born to a mother who was known to be resident within the defined geographic regions of the registers. Inclusion of only those cases that were flagged successfully ensured that all deaths and emigrations were reported, and inclusion of only those infants who were alive at age 2 years recognizes that children who have CP and die before age 2 years are underreported[17].

Classification of Covariates

Gestational age was classified into 2 groups: preterm (<37 weeks) and term infants (≥37 weeks). For birth weight, 2 classification groups were used: low birth weight (<2500 g) and normal birth weight (≥2500 g). The Carstairs index of deprivation[18] was used as a measure of socioeconomic status, using ward of residence of the mother at the time of the birth, and standardized to the U.K. population. Quintiles range from 1, the most affluent, to 5, the most deprived, and the dichotomy "deprived" (Carstairs 4 and 5) and "affluent" (Carstairs 1, 2, and 3) was used here.

The binary categories "severe" and "not severe" were used to classify impairment variables. For lower limb function, a severe impairment indicates that the child is unable to walk even with aids, uses a wheelchair, or is bedridden. For upper limb function, a severe impairment indicates that the child is unable to feed or self-dress. A severe visual impairment is defined as vision in the better eye <6/60. A severe hearing impairment is defined as a hearing loss in the better ear >70 decibels averaged across frequencies 0.5 to 4 kHz. A severe intellectual impairment is defined as an IQ <50. For the sensory and cognitive variables, assessment of impairments was made by formal testing procedures or by clinical judgment when formal testing was not possible.

Statistical Analyses

Kaplan-Meier survival estimates at ages 5 to 20 years (where data permit) by 5-year intervals were provided for each region and decade of birth for male and female individuals. Numbers of cases in each group at the beginning of follow-up and standard error at 20 years (or last year of follow-up) were also provided. Regional variations by selected birth characteristics, impairments, and severity (except hearing, for which there are insufficient deaths in those with severe impairment to make any reasonable

TABLE 1. Demographic Characteristics of Children With CP by the 5 Regions

	Mersey	North England	North Ireland	Oxford	Scotland
Notifications					
Cases	768	745	885	955	654
Deaths	90	54	67	63	51
Decade of birth					
1980s	768	198	550	494	654
1990s		547	335	461	
Gender					
Male	428	459	485	554	375
Female	340	286	400	401	279
M:F ratio	1.3	1.6	1.2	1.4	1.3

evaluations) were evaluated at 10 and 20 years. Differences between groups were evaluated using the log-rank test.[19] Cox proportional hazards models[19] were used to evaluate regional variations after adjustment for impairments, birth characteristics, socioeconomic status, and year of birth. Assumptions of proportionality were assessed by the Grambsch and Therneau methods, and best fits were evaluated using Akaike's information criterion.[20]

Expected survival proportions for the U.K. population, matched to the CP cohort by gender, year of birth, and alive at age 2 years, permitted comparisons with the general population. Expected survival estimates were computed using the relative survival package *surv2*[21] and using U.K. life tables (source: Government Actuary's Department).

RESULTS

Among the 4007 children who had early-impairment CP, whose mother was resident in the defined regions at the time of the birth, who were flagged with the relevant births and deaths register, and who were alive at age 2 years, 325 deaths were reported (Table 1). There were more male than female individuals with CP across all the regions. The ratio of male to female individuals varied from 1.2 (Northern Ireland) to 1.6 (North of England), although these were not significantly different.

In general, more children were born at term and with normal birth weight in all regions than were born preterm or with low birth weight (Table 2). Proportions in the low birth weight group varied between 43% (North of England, 1990s) and 51% (Scotland, 1980s). A similar pattern was seen in the preterm births, which ranged from 39% (the North of

TABLE 2. Secular and Regional Variations in Percentages of Children Who Were Born With Low Birth Weight, Preterm, and in Deprived Areas

	Mersey	North England	North Ireland	Oxford	Scotland
N	768	745	885	955	654
Low birth weight (≤2500 g)					
1980s	48 (0)	44 (0)	47 (1)	45 (0)	51 (0)
1990s		43 (0)	45 (5)	48 (0)	
Preterm (≤37 wk)					
1980s	47 (1)	39 (0)	44 (2)	42 (0)	48 (1)
1990s		43 (0)	45 (3)	47 (0)	
Deprived Carstairs (quintiles 4 and 5)					
1980s	67 (5)	76 (6)	79 (1)	33 (2)	90 (12)
1990s		76 (19)	79 (1)	32 (8)	

Percentages of cases for which the information on this variable is missing are in parentheses.

TABLE 3. Secular and Regional Variations in Percentages of Children With Severe Impairment

	Mersey	North England	North Ireland	Oxford	Scotland
N	768	745	885	955	654
Severe lower limb impairment					
1980s	27 (0)	30 (9)	30 (3)	33 (1)	36 (4)
1990s		27 (3)	23 (7)	33 (4)	
Severe upper limb impairment					
1980s	22 (0)	25 (6)	22 (5)	22 (2)	25 (4)
1990s		25 (3)	16 (12)	20 (3)	
Severe intellectual impairment					
1980s	30 (1)	25 (5)	33 (8)	30 (13)	3 (63)
1990s		24 (3)	22 (14)	21 (17)	
Severe visual impairment					
1980s	11 (9)	5 (11)	11 (9)	13 (7)	18 (41)
1990s		6 (4)	9 (8)	10 (7)	
Severe hearing impairment					
1980s	2 (1)	3 (9)	2 (8)	2 (10)	2 (9)
1990s		5 (3)	2 (7)	3 (3)	

Percentages of cases for which the information on this variable is missing are in parentheses.

England, 1980s) to 48% (Scotland, 1980s). The majority of the children were born in deprived areas in all regions except Oxford, where just 33% were. In Scotland, at the other extreme, >90% of the children were born in deprived areas. The proportion of cases with missing information on gestational age and birth weight was very low in all regions (<5%). Proportions of cases with missing information on socioeconomic status were higher (eg, 19% in the North of England in the 1990s).

Some variation was observed between the regions in the proportion of children with severe impairment (Table 3). For example, proportions of children with a severe lower limb impairment varied from 27% in Mersey to 36% in Scotland in the 1980s; proportions with a severe intellectual impairment varied from 25% in the North of England to ~30% in the other regions in the 1980s (omitting Scotland because 63% of data were missing). Rates of missing data were generally greater for the sensory variables (at ~10%) than motor variables (at ~5%) and were the highest and most variable for the intellectual impairment

variable (as much as 63% in Scotland but as little as 1% in Mersey). In Oxford and Northern Ireland, the proportion of cases with missing information on intellectual impairment increased in the 1990s. The rate of missing data from the North of England was lower in the 1990s than in the 1980s.

Survival From Age Two Years

The proportion of children who were alive at age 2 years and who survived to 20 years of age varied between 85% (Mersey) and 94% (North of England) for male individuals and between 87% (Mersey and the North of England) and 90% (Northern Ireland and Oxford) for female individuals (Table 4). No significant variations in these survival proportions were detected. No significant differences were observed between the genders, either within regions or across all regions combined. For comparison and on the basis of the U.K. life tables matched to the UKCP cohort by year of birth and gender and alive at age 2 years, the 20-year survival rate in the general population was 99.2% for male individuals and 99.6% for female individuals.

In the 1980s, survival of children who were born preterm was significantly better ($P < .01$) than those who were born at term; similarly, survival for those who were born with low birth weight was better than for those who were born with normal birth weight ($P < .01$), across all regions (Table 5). There was a significant regional variation for infants who were born preterm ($P < .01$) or with low birth weight ($P < .01$), with Mersey having the poorer outlook and the North of England and Scotland faring better. No significant regional variation was observed for children who were born at term or with a normal birth weight. Considering all children who were born in deprived areas, there was a significant regional variation in survival ($P < .1$), with Mersey having a poorer outlook. There was a trend, although not significant, for affected children who were born in the more affluent areas to have better survival. In contrast, in Oxford, better survival was observed in the children from the less affluent areas.

For children with a particular impairment recorded as severe, those who were born in Mersey had a poorer outlook, whereas those who were born

TABLE 4. Survival of Those Who Were Alive at Age Two Years by Region and Decade: Percentages Surviving to Ages 5, 10, 15, and 20 Years

	Male						Female					
	5 y	10 y	15 y	20 y	n	SE	5 y	10 y	15 y	20 y	n	SE
1980s												
Mersey	97	93	90	85	428	1.8	96	94	92	87	340	1.8
North England	99	98	95	94	123	2.0	97	93	91	87	75	3.3
North Ireland	96	95	93	87	296	2.7	98	96	93	90	254	2.3
Oxford	98	94	91	89	285	2.5	96	93	90	90	209	2.0
Scotland	99	96	94	93	375	1.7	99	96	93	88	279	1.7
1990s												
North England	97	93			336	1.6	98	94			211	1.6
North Ireland	98	96			189	2.3	99	97			146	2.1
Oxford	98	97			266	1.6	99	98			192	1.8

n indicates number of cases at beginning of follow-up; SE, standard error at last year of follow-up.

TABLE 5. Survival of Those Who Were Alive at Age Two Years by Selected Birth Characteristics in the 1980s: Percentages Surviving to Ages 10 and 20 Years

	10 y	20 y	n	SE	10 y	20 y	n	SE
	Normal birth weight (2500 g)				Low Birth Weight (≤2500 g)			
Mersey	94	87	400	1.7	93	89	368	1.6
North England	95	87	110	3.3	99	96	88	2.0
North Ireland	95	87	286	2.2	95	91	257	1.9
Oxford	92	87	269	2.1	97	92	225	2.4
Scotland	93	87	317	2.1	98	95	334	2.7
	Term births (37 wk)				Preterm (≤37 wk)			
Mersey	93	86	408	1.8	94	90	355	1.7
North England	95	88	120	3.1	99	97	78	1.8
North Ireland	94	87	299	2.3	96	91	238	2.0
Oxford	91	86	286	2.3	99	95	311	2.6
Scotland	93	87	337	2.1	99	95	311	2.6
	Affluent (quintiles 1 to 3)				Deprived (quintiles 4 and 5)			
Mersey	95	92	242	1.8	92	86	490	1.8
North England	96	91	45	4.2	97	92	142	2.5
North Ireland	96	90	112	3.1	95	88	434	2.5
Oxford	93	89	324	1.8	95	92	160	2.2
Scotland	96	96	56	2.5	95	90	522	1.9

n indicates number of cases at beginning of follow-up; SE, standard error at last year of follow-up.

in the North of England and Scotland had the best survival in the 1980s (Table 6). For example, in the North of England, for those with a severe lower limb impairment, 85% survived to 20 years of age, compared with just 58% in Mersey. The IQ impairment variable showed the least disparity between regions and severe visual impairment the most. Significant differences ($P < .01$) between the regions were detected for all impairment variables except for intellectual impairment. In the 1990s, of the 3 regions with data available (North of England, Northern Ireland, and Oxford), the North of England had the poorest outlook and Oxford the best for all severe impairment categories, although only significantly so ($P < .01$) for those with a severe lower limb impairment. The 10-year survival in the Oxford area improved in the 1990s, compared with the 1980s ($P < .05$), whereas in the North of England, the opposite trend was seen (although not significant). There was no change in the 10-year survival in Northern Ireland during the 2 decades.

For each impairment, the survival for children without a severe impairment ranged from 97% to 99% at 20 years, with no significant temporal or regional variations; the exception was those with a nonsevere visual impairment in the North of England in the 1990s, who had a survival rate of 95% at 10 years, significantly lower than that in the other regions ($P < .01$). Survival for the unknown severity groups showed large variations between the regions

TABLE 6. Survival of Those Who Were Alive at Age Two Years by Impairment Variables, Region, and Decade: Percentages Surviving to Ages 10 and 20 Years

	1980s				1990s			Unknown Severity		
	10 y	20 y	n	SE	10 y	n	SE	20 y	n	SE
Severe lower limb function										
Mersey	78	58	208	3.7				100	1	
North England	95	85	55	5.1	78	161	3.3	65	17	11.6
North Ireland	85	65	158	4.2	85	73	4.5	88	25	6.5
Oxford	83	71	163	4.5	93	160	2.1	50	6	20.4
Scotland	95	80	229	4.5				44	25	9.9
Severe upper limb function										
Mersey	74	51	167	4.2				100	1	
North England	94	80	47	6.1	80	157	3.5	67	12	13.6
North Ireland	83	58	117	3.5	82	47	6.0	79	34	7.2
Oxford	76	60	104	5.8	90	101	3.1	50	28	9.5
Scotland	92	70	157	6.4						
Severe intellectual impairment										
Mersey	81	63	228	3.4				40	5	21.9
North England	85	70	41	7.3	79	143	3.6	89	9	10.5
North Ireland	86	68	165	4.1	86	64	4.6	92	56	4.2
Oxford	80	67	128	4.9	89	93	3.4	90	62	37.5
Severe visual impairment										
Mersey	66	39	74	6.5				61	68	6.3
North England	89	61	9	18.1	67	32	8.7	73	22	9.5
North Ireland	84	60	56	7.1	80	27	8.3	78	53	5.9
Oxford	79	56	62	10.1	82	48	6.0	79	33	7.1
Scotland	90	72	69	5.7				92	267	1.7

n indicates number of cases at beginning of follow-up; SE, standard error at last year of follow-up. For unknown severity, results are for the 1980s cohort only.

but also had less precision, reflecting the smaller numbers of cases (Table 6).

Multivariate Survival

There was a reduction in life expectancy with increasing numbers of multiple severe impairments (Fig 1). Information was available on all 4 impairment variables for 2853 children in all of the regions (excluding Scotland because of poor intellectual impairment data) for both decades. The 475 (17%) who had either 3 or 4 impairment indicators that were classified as severe accounted for 84% of the deaths. For children with 2 severe impairments, the proportion who survived to 20 years of age was >90%, whereas with 4 severe impairments, the proportion who survived to 20 years of age was just 40%. Considering children with all 4 impairment variables recorded as severe, no significant regional variation was observed among the 4 regions. However, once again, Mersey had a slightly poorer outlook and Oxford slightly better (32% vs 42% at 20 years).

When a multivariate proportional hazards model was fitted, the best predictors of survival (ie, with the largest estimated relative hazards from age 2 to 20 years between the severe and not severe) were in descending order: severe lower limb impairment (relative hazard: 5.01; 95% confidence interval [CI]: 2.5–10.1), severe intellectual impairment (relative hazard: 3.91; 95% CI: 2.3–6.8), severe upper limb impairment (relative hazard: 3.10; 95% CI: 1.8–5.5), and severe visual impairment (relative hazard: 2.12; 95% CI: 1.6–2.9).

Furthermore, a significant interaction was found between deprivation status and birth weight: for those who were born in an affluent area, the effect of birth weight was 2.2-fold (95% CI: 1.2–4.1) that of the effect of birth weight for those who were born in a deprived area (Fig 2). In affluent areas, infants of normal birth weight had an increased relative hazard of 2.0 (95% CI: 0.8–5.4) compared with those of low birth weight, whereas in deprived areas, infants of normal birth weight had a decreased relative hazard of 0.93 (95% CI: 0.7–1.3) compared with those of low birth weight. For those who were born in an affluent area with normal birth weight and with all 4 impairment variables recorded as severe, <30% survive to 20 years of age, yet for those who were born in affluent areas and had low birth weight and all 4 impairment variables recorded as severe, >50% survived to 20 years of age. After the inclusion of these variables in the multivariate model, region or year of birth did not make a significant additional contribution.

DISCUSSION

Findings

We have shown that after adjustment for severity of impairment, children who had CP, were born in affluent areas, and had a low birth weight fare much better than children who were born in a similar area had a normal birth weight. However, this result was not replicated (and may even have been reversed) among those who were born in deprived areas. Significant regional variations were evident when the survival by a particular severe impairment variable
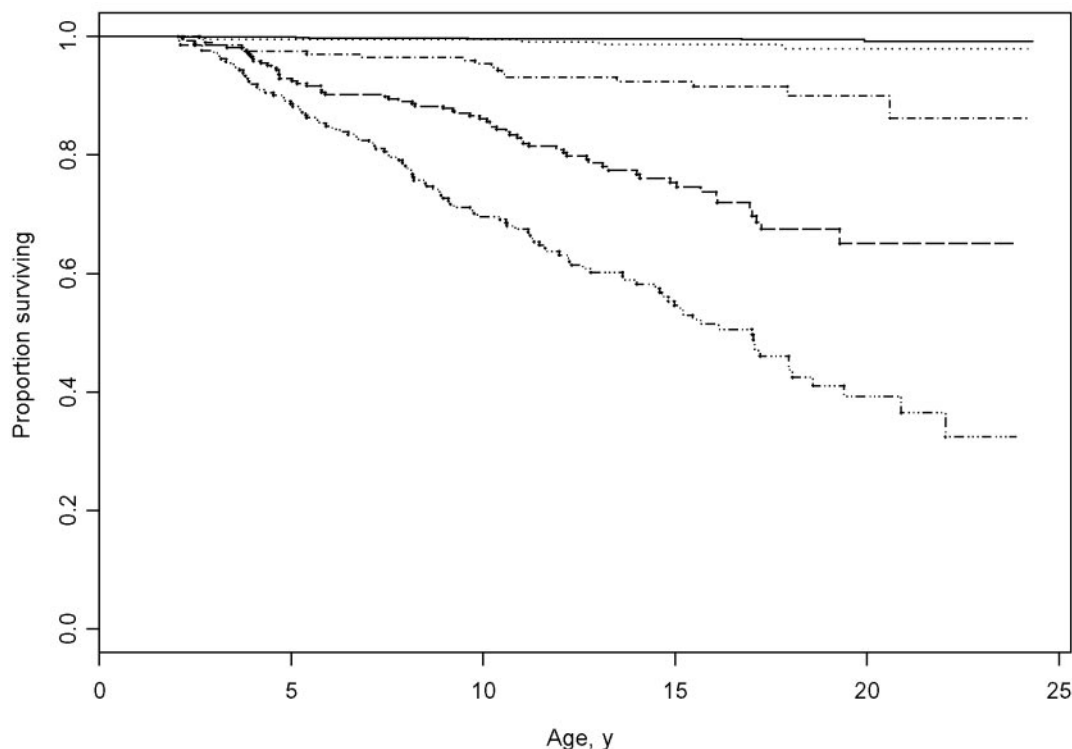

Fig 1. Survival of those who were alive at age 2 years by number of severe impairments for all regions combined: —, no severe impairments, 1843 children with 10 deaths; · · · · · · , 1 severe impairment, 341 children with 5 deaths; – · –, 2 severe impairments, 194 children with 17 deaths; — —, 3 severe impairments, 265 children with 65 deaths; – · · · –, 4 severe impairments, 211 children with 105 deaths.
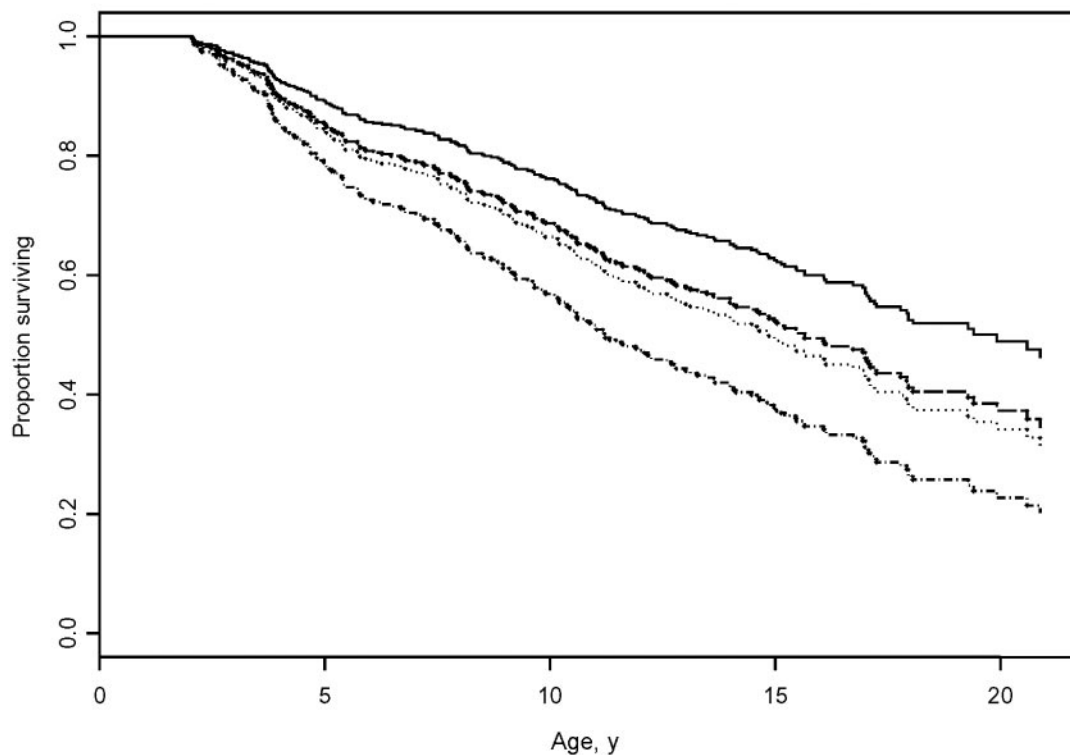
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Fig 2. Predicted survival for those who were alive at age 2 years and had 4 severe impairments by birth weight and socioeconomic status, for all regions combined: —, low birth weight and affluent; — —, normal birth weight and deprived; · · · · · · , low birth weight and deprived; – · –, normal birth weight and affluent.

was compared. However, no regional variations exist after adjustment for multiple impairments, birth characteristics, and socioeconomic status. Furthermore, within our multivariate analysis over the 2 decades, we found no evidence of any secular changes in survival.

Of those who had 4 severe impairments and were born with normal birth weight and in affluent areas, <30% will survive to 20 years of age. This estimate of survival for the most severely impaired group is worse than that previously published[9] (although within expected margins of error) and is likely to be attributable to the additional contribution that socioeconomic status and birth weight make to survival outlook.

Birth Characteristics

The survival of those who were born with a low birth weight, contrary to what may be expected, although previously reported,[9] is better than that of children who were born with a normal birth weight. Infants who are born preterm or with low birth weight are less likely to have severe impairment than those who are born at term or with normal birth weight; even after adjustment for level of impairment, children who are born preterm or with low birth weight still have better survival.[9] However, our research shows that this holds true only for those who were born to mothers from affluent areas and that little difference is evident between the birth weight groups for those who were born to mothers from deprived areas. Furthermore, for those who were born to mothers from affluent areas, the 2-fold

difference in risk reported here between those who were born with a normal birth weight and those who were born with a low birth weight is greater than that previously reported.

Although we have not presented our data on children with late-impairment CP because we know that it is underascertained, it has been shown[9] that children with late-impairment CP have a poorer outlook than those with early-impairment CP for a particular severe impairment.[9] If one considers gestational age to be a surrogate measure for age at insult and continues this time axis beyond birth to age at insult for late-impairment CP, then it could be hypothesized (again conditioning on severity of impairment) that there is a decrease in life expectancy with increasing age of insult. It would be of additional interest to evaluate influences of socioeconomic status on such outcomes.

Socioeconomic Variations

All of the UKCP regions except for Oxford happen to cover the more deprived regions of United Kingdom.[22] A substantial class gradient is known to exist in childhood mortality. Using the National Statistics Socio-Economic Classification, infant mortality rates in the semiroutine and routine laborers (the 2 lowest occupational bands of the National Statistics Socio-Economic Classification) are 5.9 to 6.4 per 1000 live births compared with just 1.8 to 2.5 per 1000 live births in the higher professions.[23] Perinatal, infant, and childhood mortality also vary by region, much of which might be associated with variations in social class.[24,25]

Socioeconomic variations may have wide ranging implications. First, differences in perinatal survival between the socioeconomic groups may affect underlying distributions of CP between the regions, affecting infants who survive to the age 2 threshold and may be ascertained in a study of this nature. Second, socioeconomic status is related to birth weight (with more infants from lower socioeconomic bands being born with low birth weights). A so-called low birth weight paradox exists where infants who were born with a low birth weight have a lower perinatal mortality when born to women of lower socioeconomic status than a higher socioeconomic status.[26] However, survival for those children with CP would suggest a paradox of the normal birth weight infants: children who have CP and are born with normal birth weight have a lower mortality when born in deprived areas than in affluent areas.

Regional Variation and Data Ascertainment

Some of the disparity in international comparisons of survival of CP has been attributed to differences in ascertainment of the very mild and very severe impairment.[10] Any assessment of regional variation must depend to some extent on assumptions of complete ascertainment or at least equivalent patterns of ascertainment across the regions. Exclusion of children who do not live to age 2 years limits the effect of underascertainment of severe cases. Focusing on those with severe impairment limits the effect of underascertainment of the very mild cases.

An investigation of possible underascertainment using capture-recapture methods in the Oxford region revealed that it was likely that between 13% and 21% of cases had not been ascertained across all years of birth and that this rate may be higher in more recent years (A. Silva, PhD, personal communication, 2004). Thus, the observed improvement in survival in the 1990s for Oxford could potentially be confounded by underascertainment issues. However, the estimated degree of underreporting in Oxford was also found to be lower for those with severe impairment compared with those with less severe impairment, and so the improvement in survival observed in those with severe impairment in Oxford in the 1990s may well represent a real improvement.

Ascertainment may also be influenced by whether data were collected retrospectively or prospectively. Retrospective methods may be less likely to ascertain children who had severe impairment and have already died. For instance, a more favorable outlook was observed in the North of England 1980s cohort than the 1990s. The survey initially obtained its cases by retrospective periodic searches, in 1985, 1990, and 1995, and then subsequently by prospective methods.

Implications of Potential Variations in Recording and Definitions

In an attempt to avoid some of the variation as a result of definition and classification of impairments, we considered only the binary classification of severe or not severe. However, variations in recording of severe impairments may account for some of the variability observed in survival between the regions for those who have severe impairment. In particular, children for whom severity status is unknown may be more likely to have severe impairment than not (eg, because of an inability to test the degree of severity, or the child's dying before testing was conducted). The Mersey register distinguishes between children for whom visual impairment is unknown and those for whom it is impossible to test. The survival for those for whom it is impossible to test is similar to those who have severe impairment.[9] The North of England register also makes this distinction, but the 3 other registers do not. A detailed statistical analysis (not shown here) showed that such missing data were unlikely to alter the overall conclusions drawn (namely, that there exists regional variation when considering single severe impairments but not when considering multiple severe impairments).

Generalizability

Large cohorts of children are needed to derive precise estimates of life expectancy for children with CP. A known underreporting of CP on death certificates prevents such information from being taken from routine statistics. This large collaborative study helps to overcome issues of power and small numbers when subgroups of children with CP are studied. The geographically defined populations of each of the registers allows estimates to be applied to total populations.

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Krug N. *New York Times*. September 25, 2005

Noted by JFL, MD

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